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Contents

Original Papers

Protocols

Viewpoint

Patient Recruitment 2.0: Become a Partner in the Patient Journey Using Digital Media (e17)

Michael Lindemann, Tobe Freeman, Timothy Kilchenmann, Shuree Harrison, Margaret Chan, Mark Wygonik, Lea Haines, Christian Gossens.
4 5 4

Original Paper

Can Internet-Based Sexual Health Services Increase Diagnoses of Sexually Transmitted Infections (STI)? Protocol for a Randomized Evaluation of an Internet-Based STI Testing and Results Service

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Abstract

Background: Ensuring rapid access to high quality sexual health services is a key public health objective, both in the United Kingdom and internationally. Internet-based testing services for sexually transmitted infections (STIs) are considered to be a promising way to achieve this goal. This study will evaluate a nascent online STI testing and results service in South East London, delivered alongside standard face-to-face STI testing services.

Objective: The aim of this study is to establish whether an online testing and results services can (1) increase diagnoses of STIs and (2) increase uptake of STI testing, when delivered alongside standard face-to-face STI testing services.

Methods: This is a single-blind randomized controlled trial. We will recruit 3000 participants who meet the following eligibility criteria: 16-30 years of age, resident in the London boroughs of Lambeth and Southwark, having at least one sexual partner in the last 12 months, having access to the Internet and willing to take an STI test. People unable to provide informed consent and unable to read and understand English (the websites will be in English) will be excluded. Baseline data will be collected at enrolment. This includes participant contact details, demographic data (date of birth, gender, ethnicity, and sexual orientation), and sexual health behaviors (last STI test, service used at last STI test and number of sexual partners in the last 12 months). Once enrolled, participants will be randomly allocated either (1) to an online STI testing and results service (Sexual Health 24) offering postal self-administered STI kits for chlamydia, gonorrhoea, syphilis, and HIV; results via text message (short message service, SMS), except positive results for HIV, which will be delivered by phone; and direct referrals to local clinics for treatment or (2) to a conventional sexual health information website with signposting to local clinic-based sexual health services. Participants will be free to use any other interventions or services during the trial period. At 6 weeks from randomization we will collect

self-reported follow-up data on service use, STI tests and results, treatment prescribed, and acceptability of STI testing services. We will also collect objective data from participating STI testing services on uptake of STI testing, STI diagnoses and treatment. We hypothesise that uptake of STI testing and STI diagnoses will be higher in the intervention arm. Our hypothesis is based on the assumption that the intervention is less time-consuming, more convenient, more private, and incur less stigma and embarrassment than face-to-face STI testing pathways. The primary outcome measure is diagnosis of any STI at 6 weeks from randomization and our co-primary outcome is completion of any STI test at 6 weeks from randomization. We define completion of a test, as samples returned, processed, and results delivered to the intervention and/or clinic settings. We will use risk ratios to calculate the effect of the intervention on our primary outcomes with 95% confidence intervals. All analyses will be based on the intention-to-treat (ITT) principle.

Results: This study is funded by Guy's and St Thomas' Charity and it has received ethical approval from NRES Committee London-Camberwell St Giles (Ref 14/LO/1477). Research and Development approval has been obtained from Kings College Hospital NHS Foundation Trust and Guy's and St Thomas' NHS Foundation Trust. Results are expected in June 2016.

Conclusions: This study will provide evidence on the effectiveness of an online STI testing and results service in South East London. Our findings may also be generalizable to similar populations in the United Kingdom.

Trial Registration: International Standard Randomized Controlled Trial Number (ISRCTN): 13354298; http://www.isrctn.com/ISRCTN13354298 (Archived by WebCite at http://www.webcitation.org/6d9xT2bPj)

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KEYWORDS

sexually transmitted infections; eHealth; Internet; services

Introduction

Sexually transmitted infections (STIs) are an important cause of morbidity and mortality worldwide, and a key indicator of sexual ill health. Globally, incident cases of curable STIs (chlamydia, gonorrhoea, syphilis, and trichomonas vaginalis) rose from 448.3 million in 2005 to 498.9 million in 2008 [1]. While global incidence rates of HIV infection are on the decline, prevalence remains significant: UNAIDS estimated 2.3 million new HIV infections in 2012 and that 35.3 million people were living with HIV [2]. Moreover, HIV/AIDs represents 3.4% of the total global disease burden and is the seventh leading cause of all disability-adjusted life years worldwide [3].

In England, 448,422 new STI diagnoses were made in 2012, with higher rates recorded among young heterosexuals, men who have sex with men, and some black and ethnic minority groups [4]. Importantly, the patterning of STIs reflects stark health inequalities. A recent probability sample survey found that men and women from the most deprived areas of Britain had greater odds of testing positive for chlamydia (the most common STI) than those from wealthier areas (men, adjusted OR 3.42 (95%CI, 1.28-9.16), *P*=.003; women, adjusted OR 4.01(95%CI, 1.67-9.63), *P*=.008) [5].

Increasing diagnoses of untreated STIs is a key public health objective, both in the United Kingdom and internationally, to reduce onward transmission of infection and prevent long term health complications [6]. Increasing access to HIV testing and ensuring early diagnoses of HIV in high prevalent areas is of particular concern. In 2013, in the United Kingdom, it was estimated that a quarter of those living with HIV were unaware that they were infected and 42% of those diagnosed with HIV were diagnosed late, after the point at which treatment should have begun [7].

Internet-based STI testing is considered a promising means for increasing access to STI testing and reaching high-risk groups [8]. There are a number of models for Internet-based STI testing. For the purposes of this study we focus on a model which enables users to order a test kit online, take self-administered samples in their home, return test samples to a laboratory and receive their results via SMS text messaging (short message service, SMS), email or phone call. As far as we are aware, there have been no experimental studies to demonstrate the effectiveness of Internet-based testing services compared to standard face-to-face clinical pathways.

Observational studies have been encouraging, reporting high STI positivity among service users and reaching populations with a combination of both sociodemographic and behavioral risk factors. In the United States, Ladd and colleagues found that out of 205 rectal samples ordered and returned by women using the "iwantthekit" website between January 2009 and February 2011, 18.5% were positive for at least one STI [9]. The majority of women in the sample were single (91.2%), young (mean age 25.8 years) and of African-American ethnicity (50.0%). Half had never used condoms for rectal sex (48.7%). A study with male users of the same website found that of 501 STI kits returned by men over the age of 14, between September 2006 and May 2009, 21% tested positive for chlamydia, gonorrhoea or trichomonas vaginalis [10]. The majority of users were young (median age 24.5 years), single (84%), and either white (47%) or black ethnicity (45%). While these studies are promising, sample sizes have been small and no comparison has been made with users of face-to-face pathways.

In the United Kingdom there is limited evidence on Internet-based STI testing. One descriptive study reported trends in chlamydia testing among 15-24 year olds. It found that Internet tests, which have not been widely promoted, represented 5% of all tests within the National Chlamydia Screening Programme (NCSP) between 2006 and 2010 [11]. A higher proportion of Internet tests were positive compared to tests conducted in general practice services (7.6% vs 5.6%) but

slightly lower than in community-based sexual and reproductive health services (7.6% vs 8.2%). Compared to testers in face-to-face settings, a higher proportion of Internet testers were men, of white ethnicity, and in the upper age group (20-24 years).

Recent exploratory qualitative studies with young people in the United Kingdom suggest that Internet or mobile applications of sexual health services are likely to be acceptable to this population, due to ease and convenience. Privacy, trust and credibility of websites or apps were highlighted as important considerations for service development [12,13]. However, there is some uncertainty on whether Internet services can reach marginalized populations. In Scotland, Lorimer and McDaid [14] found that young men from more deprived areas seemed more disconnected from Internet technology and stated a preference for face-to-face services. This echoes findings from analysis of chlamydia testing data from England (discussed earlier), which found that a higher proportion of tests conducted in face-to-face services (GP and SRH clinics) were from more deprived areas, compared to Internet tests [11].

Given the limited evidence base it is difficult to draw conclusions on the effectiveness of Internet-based pathways in diagnosing a range of STIs compared to face-to-face service pathways.

Sexual Health 24 (SH:24) is an innovative Internet-based sexual health service that aims to improve access to sexual health services in the London boroughs of Southwark and Lambeth by addressing both supply and demand side barriers to care. Southwark and Lambeth have some of the highest rates of STIs in England, as well as high rates of teenage pregnancy and abortion [15]. Current face-to-face services are unable to meet demand with the result that many people are turned away from services.

In November 2014, SH:24 launched its first online product (minimal viable product 1) - an online STI testing and results service allowing users to order free postal STI kits, receive their results by SMS text messaging (or by phone in the event of a positive HIV result), and be referred on to specialist sexual health clinics for treatment. Over the course of this 4 year project, SH:24 will build in complexity, gradually adding increasing layers of functionality to the website, such as telephone support services and contraceptive services. SH:24 will be fully embedded within local sexual health economies ensuring that care pathways are integrated with clinical and other local services: for example by ensuring that users with acute STIs are signposted to face-to-face clinical care.

As an untested intervention, SH:24 has implications for the commissioning of sexual health services not only in London but also nationally. In line with national and international quality frameworks, SH:24 will be evaluated using a variety of data sources and methodologies to assess whether it delivers a safe, effective, patient-centered, timely, efficient, and equitable service [16,17]. This study will focus on establishing whether SH24 delivers an effective Internet-based STI testing and results service compared to face-to-face STI testing services, thus contributing to the international evidence base in this field.

Methods

Study Design

We will carry out a randomized controlled trial (Figure 1). Participants will be randomly allocated either (1) to a sexual health website (SH:24) offering free postal STI kits for chlamydia, gonorrhoea, syphilis and HIV, results via SMS text messaging (positive results for HIV will be delivered by phone), and direct referrals to clinic-based treatment options; or (2) to a conventional sexual health information website with signposting to local clinic-based sexual health services. Participants will be free to use any other service or intervention during the trial period.

Eligibility

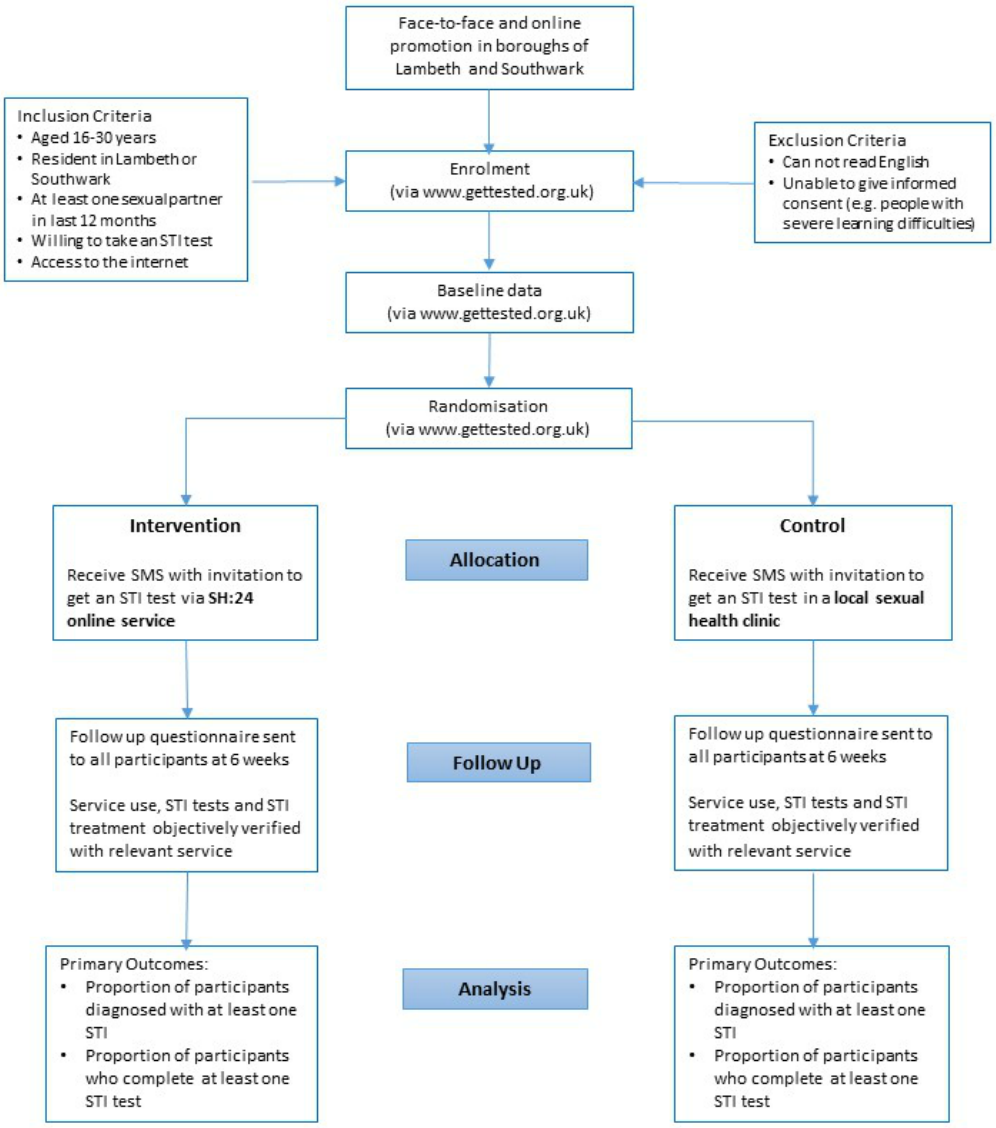
Participants will be eligible if they are between 16 and 30 years of age, resident in the London boroughs of Lambeth and Southwark, sexually active (at least one sexual partner in the last 12 months), willing to take an STI test, and have access to the Internet (owner of a mobile phone or able to access a laptop, tablet, personal computer in their own home). Participants will be excluded if they are unable to read in English as the websites will be in English. Those unable to give informed consent, such as people with severe learning difficulties, will also be excluded.

Recruitment and Consent

This study is being conducted in community settings in the boroughs of Lambeth and Southwark in South East London. The study coordinator, together with a team of research assistants, will approach community networks, organizations, and institutions such as further education colleges, universities, patient groups, sexual health advocacy groups, sports centers, entertainment and leisure venues and major employers to recruit participants. We will also utilize social media sites popular among our study population. These will include Facebook, Twitter, and dating applications for gay men such as Scruff and Grindr.

After potential participants have been assessed for their eligibility, they will be provided with detailed verbal and written information about the study, and given the opportunity to ask any questions. If the participant agrees to participate, we will ask them to provide consent via the trial website (eg, using a mobile phone) or via paper-based forms. If potential participants would like more time to consider their involvement, we will give them the contact details of the study coordinator so that they can talk through any queries or doubts. Potential participants will also be able to access the study website independently, for example via social media sites.

Figure 1. Consolidated Standards of Reporting Trials (CONSORT) flow chart.



Allocation

Participants will be asked to enter baseline data directly onto the trial website or using paper-based forms. After baseline information has been submitted, an independent computer-based randomization program will generate a unique research number and allocate participants to either the intervention or control group. Participants will be sent a SMS text message with the URL of their allocation.

We will allocate by minimization, taking into account gender (male, female), age (16-19, 20-24, 25-30), number of sexual partners (1, 2+) and sexual orientation (men who have sex with men [MSM], all other groups), where all factors have equal weight in determining marginal imbalance. To introduce a random element, allocation will be weighted toward the underrepresented group using a probability of .8. In all other instances, participants will be allocated in a 1:1 ratio.

Due to the nature of the intervention, participants will know to which arm they have been allocated. Laboratory staff carrying out STI tests will process the tests as per routine care, and will be unaware that samples are linked to a randomized controlled trial. Researchers assessing the outcomes will be blind to the treatment allocation.

Intervention Group

Through a design-led and user-centered approach, SH:24 has created an appropriate Internet-based sexual health service with the aim of improving access to sexual health services in the boroughs of Southwark and Lambeth.

Participants in the intervention group will be directed to a website which will offer them: sexual health risk assessment; the opportunity to order self-administered sample collection kits for HIV, chlamydia, syphilis, gonorrhoea; results given by SMS text message and by phone (positive HIV result only); direct referral to local clinics for treatment; health promotion information; signposting to clinic and other social services.

Participants who test positive for chlamydia, syphilis or gonorrhoea infection will be sent a SMS text message with their result and details of local sexual health clinics where they will be able to obtain treatment. Participants who test positive for HIV will be informed by phone by a health professional.

Control Group

Participants in the control group will be directed to a sexual health information website with signposting to clinic-based services. These include the Camberwell Sexual Health Centre, the Burrell Street Sexual Health Centre, the Lloyd Clinic and a range of local community-based sexual and reproductive health services in Lambeth and Southwark, where they can obtain an STI test in person. The information website will provide the address, contact details, and location of the clinics (via a google map image), alongside the URL link to the clinic website.

All participants will be free to use any other services or interventions during the trial period.

Outcomes

Our primary outcomes will be the proportion of participants diagnosed with at least one STI in each arm at 6 weeks from randomization, and the proportion of participants who complete at least one STI test in each arm at 6 weeks from randomization. The secondary outcomes are listed in Textbox 1.

Textbox 1. Secondary outcomes.

- The proportion of STI tests that are positive in each arm
- The proportion of participants who are prescribed treatment in each arm
- Time from randomization to completion of an STI test in each arm
- Time from randomization to treatment in each arm
- Time from diagnosis to treatment in each arm
- The proportion of the intervention group who agree that Internet-based STI testing is acceptable
- The proportion of the intervention group who adhere to the prescribed Internet-based testing pathway, without seeking additional support from face-to-face services

Data Collection

Eligibility data will include age, postal code, independent access to the Internet (owner of mobile phone or access to tablet, laptop or computer at home), at least one sexual partner in the last 12 months, willingness to take an STI test.

Participants will be asked to enter their baseline data directly onto the trial website or using paper-based forms. We will collect the following: contact details including first name, surname, main mobile number, email address, and primary postal address; demographic data including date of birth, gender, ethnicity, and sexual orientation; sexual health behaviors including last STI test, service used at last STI test, and number of sexual partners in the last 12 months

Self-reported follow up data at 6 weeks will be collected by post or online, according to the preference of participants. We will assess whether Internet-based testing was acceptable to participants and collect data on service use, STI tests and results, and whether treatment was prescribed. We will also collect objective data on STI tests, results, and treatment from SH:24 and local sexual health services, with participants' prior consent.

Lab Processing

The Doctors Laboratory (TDL) will process all returned samples for the intervention group and the results will be captured by SH:24 data systems. Samples taken in face-to-face settings will be processed as per routine care. The study coordinator will contact the local services and SH:24 to obtain participants' test results and treatment details, with prior consent. These data will be anonymized and uploaded into the secure trial website.

Follow-Up

We will utilize evidence-based methods identified in systematic reviews to minimize losses to follow up [18]. These include providing incentives to all participants, contacting respondents prior to sending questionnaires and contacting nonresponders using phone call, texting, email, and post. The research team will verify participant addresses at enrolment or shortly after, and attempt to contact participants who have provided an incomplete or unknown address.

To maximize participation and response rates we will provide an incentive (such as a lollipop or chocolate) at enrolment and an unconditional incentive of £5 at 6 weeks when we send out the follow-up questionnaire. Participants will then receive a further £5, on receipt of their completed questionnaires. They will be informed of the incentives at enrolment. Our incentives will not exceed a monetary value of £15 per participant.

Sample Size

The study is powered for the first of our primary outcome measures which is the proportion of participants diagnosed with at least one STI in each arm. Two factors determine the number of participants needed for this trial: the estimated proportion of participants with an STI and the size of the treatment effect.

Our estimates are based on the following data:

The Greenwich sexual health service has demonstrated a 50% return rate among users who order test kits online (personal communication Dr David Pinson, Health Improvement Principal, Royal Borough of Greenwich). Eligibility for this study is restricted to people who are willing to take an STI test. However, not all of those allocated to the intervention group are likely to order a test kit. We estimate that 30% will not complete this first step. Among the 70% who order a kit, we assume that 50% will return the kit for analysis, based on the Greenwich data. Following these assumptions, 35% of the intervention group are likely to complete an STI test.

There are no available data which would give us an estimate of the likely numbers that will get tested in the control group.

However, we assume that far fewer people (10%) are likely to seek a test in clinic-based settings.

Chlamydia is the most commonly diagnosed STI of the four STIs of interest in this study both at the national level (England) and at the local authority level (Lambeth and Southwark) [4,15]. We based our prevalence estimates on the proportion of positive chlamydia tests among 15-24 year olds in general practice settings in Lambeth and Southwark, which was 6% in 2012 [19].

We based our estimated losses to follow up on previous eHealth studies in the United Kingdom which have achieved 90% follow up [20].

A sample size of 3000 would have 90% power (two-sided alpha=5%) to detect a relative risk of 3.5, (2.1% risk of diagnosis in the intervention group vs 0.6% risk of diagnosis in the control group), allowing for 10% losses to follow up. This equates to 10% of the control group being tested, with a 6% probability of infection as in general practice settings and 35% of the intervention tested with a 6% probability of infection as in general practice settings.

With regard to our co-primary outcome measure, with 3000 participants we would have 99% power (two-sided alpha=5%) to detect an absolute difference of 25% between the proportion of participants who complete a test in the intervention group versus the proportion who complete a test in the control group (35% versus 10%).

Statistical Methods

The analysis of data will adhere to the prespecified statistical analysis plan outlined below. The analyses of the co-primary outcomes are described in detail; analyses for other outcomes follow the same principles unless otherwise specified.

There are no planned interim analyses and so no rules for stopping early. Analyses comparing the interventions will follow the ITT principle as far as possible [21]. Analyses will include participants with no missing outcome data in their randomized groups. Any estimate described below will be accompanied by 95% confidence intervals.

The primary analysis of the first co-primary outcome (STI diagnosis) will estimate the proportion of STI diagnoses for the SH:24 vs conventional sexual health services via a risk ratio. Treatment allocation balances gender, age, number of sexual partners in the last 12 months, and sexual orientation, and these need to be reflected in the analysis. This will be done by weighting on the inverse propensity score (estimated by logistic regression) to reduce the variance of estimates and obtain confidence intervals of the correct width [22].

Some outcome data are expected to be missing. Missing data may occur if participants do not complete a 6-week follow-up questionnaire and attend a different sexual health service (ie, not a local clinic or SH:24) . The principle analysis will assume that the distribution of STI diagnoses among these participants is identical to those with observed data, conditional on propensity scores–missing at random–and so will be based on the weighted analysis of the complete cases. Sensitivity analyses assuming departures from missing at random will proceed via multiple imputation of outcome, using inverse probability weighting on the estimated propensity score and with allocated group as the only covariate. Assuming “missing not at random” mechanisms, the odds of STI diagnoses for missing participants will be varied to be ¼, ½, 2 and then 4 times larger than in the missing at random (MAR) analysis. This will be done in a factorial manner, separately for each arm. We judge these fractions to be reasonable, though a value of ¼ in one arm and 4 in the other (or vice versa) is at the boundary of what is plausible. The risk difference (rather than ratio), weighted by the inverse propensity score, and the proportion of STI diagnoses in each arm will also be presented. Finally, the proportion of STI tests taken that are positive will be summarized by arm, though no comparison of the groups will be given because this analysis excludes individuals based on a variable that will be heavily influenced by randomization.

The primary analysis of the second co-primary outcome (completion of an STI test) and secondary outcome (prescribed treatment) will follow the same principles as above. Again, the risk ratio, risk difference, and proportions in each arm will be reported.

The time from randomization to (1) test completion and (2) treatment are of interest. Therefore, for each measure we will estimate the restricted-mean survival time (RMST) in each arm, setting the restricted mean time t^*=6 weeks for time to test and t^*=3 months for time to treatment. This will be estimated from a “3df/1df” Royston-Parmar model and the difference in restricted-mean survival time estimated [23]. The median survival time from diagnosis to treatment in each arm will also be summarized. No comparison will be made between groups as this analysis excludes individuals based on a variable (STI diagnosis) that is heavily influenced by randomization.

For the SH:24 group, the proportion of participants who deem Internet-based testing to be acceptable will be summarized, as will the proportion who adhere to the SH:24 testing pathway.

It is possible that differences between groups will vary according to age, level of deprivation, sexuality, ethnicity, and gender. Subgroup analyses will be done for these characteristics and interaction tests will be performed. These will have low impact as we anticipate any interactions will be small. Further, these analyses will be regarded strictly as hypothesis-generating.

Ethical Arrangements

Informed Consent

All participants recruited into the trial will be provided with information about the study (online or in hard copy) and given the opportunity to ask questions and clarify queries at the time of recruitment and subsequently with the study coordinator by email or by phone. The recruiting staff will check that participants are aware that consenting to participate means that they will be encouraged to undertake an STI test.

Participants' Rights

Participants will be able to contact the trial co-ordinating center at Kings College London by email or by phone with any queries or doubts for the duration of the trial. If they request to be withdrawn from the study their status will be changed to

"withdrawn" on the study website and they will be excluded from participant lists for follow up.

Personal details will be stored on a password protected computer held on a secure server at Kings College London. This information will be stored separately from any anonymized research data, and will be deleted at the end of the study.

Participants in the intervention arm who report symptoms of STIs will be advised to see a health professional in a clinic-based setting. If they wish to continue with the Internet-testing pathway they will be allowed to do so. Participants in the intervention arm will be informed about any positive results for chlamydia, gonorrhoea, and syphilis by SMS text message and they will be texted information of local clinics where they can receive treatment. Participants who test positive for HIV will be informed by telephone by a trained health professional and will be referred on to specialist HIV services.

Participants' Safety

The intervention provides an opportunity to obtain a postal STI kit, notification of STI diagnoses, and opportunities for STI treatment. It is unlikely to cause any harmful effects. Participants who lack privacy in their home or participants who are in abusive interpersonal relationships may risk possible consequences if they participate in Internet-based STI testing. However, this risk will be minimized as we will ensure that at recruitment participants have sufficient privacy to participate in the trial. Furthermore, the website can be accessed via devices such as mobile phones and postal test kits will be sent out in packages that do not have any identifying features. Other large scale studies using Internet-based testing have not reported any related safety concerns [24]. The intervention website will also provide clear signposting to counselling services for violence as well as contraception and abortion services.

All sexual health services participating in this trial routinely deliver test results via SMS text message. There is a small risk that friends and partners may see participants' results if phones are shared. However, as in routine care, if participants are concerned about their privacy they can opt to receive their results via different methods (eg, via post). The intervention delivers the results via SMS text message (except in the case of a positive result for HIV, which is delivered via phone). However, this is made very clear on the website and participants can choose not to order an STI test kit online, if they are concerned about their privacy.

Retention of Trial Documentation

We will retain the trial documentation for 10 years.

Results

By April 21, 2015, 1405 participants were randomized. We are currently recruiting and a timeline of our trial is included in Table 1.

Table 1. Trial timeline.

Timeline	Tasks
Months 1-6 (April-September 2014)	Trial setup
By month 6 (September, 2014)	Intervention website user tested and finalized (to be developed and finalized by SH:24)
	Completed and user tested trial database and randomization system
	Recruitment strategy designed and completed
By month 16 (July, 2015)	Recruitment to the trial completed
By month 22 (January 2016)	All follow up completed
By month 24 (March, 2016)	Data cleaned, trial database closed
By month 27 (June, 2016)	Analysis of trial results completed
	Paper submitted for publication

Discussion

This trial will provide a robust and rigorous evaluation of a nascent online STI testing service in an area of South East London, characterized by poor levels of sexual health relative to the rest of the country. It will assess the added contribution of this service with respect to two distinct but interrelated outcomes: diagnoses of STIs and uptake of STI testing.

We envision that our findings will be highly policy-relevant and will be well-placed to inform decision-making for the effective commissioning and delivery of STI testing services in London. Our findings may also be generalizable to similar populations in the United Kingdom.

To our knowledge, this is the first RCT of an Internet-based testing service, which makes a direct comparison with standard face-to-face care. The findings from this study will therefore make a timely contribution to the international evidence base in this field.

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Conflicts of Interest

Dr Baraitser is a director of SH:24 which is the community interest company that holds the grant from Guy's and St Thomas' Charity to develop the intervention. Additional oversight for the evaluation is provided by the Evaluation Steering Group and an independent adviser, Dr Caroline Free, based at the London School of Hygiene and Tropical Medicine.

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Abbreviations

HIV: human immunodeficiency virus
ITT: intention-to-treat
MAR: missing at random
MSM: men who have sex with men
NCSP: National Chlamydia Screening Programme
OR: odds ratio
RMST: restricted-mean survival time
SMS: short message service
STI: sexually transmitted infection
TDL: The Doctors Laboratory

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Protocol

Computerized Tailored Interventions to Enhance Prevention and Screening for Hepatitis C Virus Among People Who Inject Drugs: Protocol for a Randomized Pilot Study

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Abstract

Background: Hepatitis C virus (HCV) infection is a growing problem among people who inject drugs. Strategies to reduce disease transmission (eg, syringe exchange programs) and facilitate HCV screening and linkage are available but are under-utilized in many communities affected by injection drug use. Novel approaches to increasing the use of these strategies are needed.

Objective: The goals of this project are to (1) develop and pilot test a computerized tailored intervention for increasing HCV screening and decreasing risky drug use behavior among people who inject drugs and (2) determine the feasibility of disseminating such an intervention using peer-based referrals in the setting of a community-based syringe exchange program.

Methods: This 2-arm, randomized pilot study is being conducted in a large-volume, multisite syringe exchange program in southern Wisconsin. A social network–based strategy was used to recruit a total of 235 adults who reported past-month injection of opioids, cocaine, or methamphetamine. Network recruiters were identified among clients requesting services from the syringe exchange program and were enlisted to refer eligible peers to the study. All participants completed a computer-adapted questionnaire eliciting information about risk behaviors and their knowledge, attitudes, and prior experiences related to HCV screening. Subjects were then randomly assigned to receive usual care, consisting of standard counseling by syringe exchange staff, or the Hep-Net intervention, which provides algorithm-based, real-time tailored feedback and recommendations for behavior change in the style of motivational interviewing. Changes in drug use behaviors and attitudes will be assessed during a second session between 90 and 180 days after the baseline visit. Frequency of repeat HCV testing and HCV incidence will be assessed through a database search 1 year after study completion.

Results: Recruitment for this study was completed in April 2015. Follow-up of enrolled participants is expected to continue until March 2016. Network recruiters were enrolled who referred a total of 195 eligible peers (overall N=235). At baseline, the median age was 34 years; 41.3% (97/235) were non-white; and 86.4% (203/235) reported predominantly injecting heroin. Most participants (161/234, 68.8%) reported sharing injection equipment in the past and of these, 30.4% (49/161) had never been tested for HCV.

Conclusions: This study will provide preliminary evidence to determine whether incorporating computerized behavioral interventions into existing prevention services at syringe exchange programs can lead to adoption of healthier behaviors.

Trial Registration: ClinicalTrials.gov NCT02474043; https://clinicaltrials.gov/ct2/show/NCT02474043 (Archived by WebCite at http://www.webcitation.org/6dbjUQG7J)

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KEYWORDS

hepatitis C; substance abuse, intravenous; needle exchange programs; health behavior

Introduction

Objectives

The overall goal of this project is to explore whether deploying a computer-adapted behavioral intervention coupled with onsite, rapid hepatitis C virus (HCV) screening is a feasible and acceptable approach to reducing transmission risk behavior and improving HCV case detection in the setting of a syringe exchange program. The intervention described in this paper incorporates lessons from formative research conducted with the target population [1] and prior experience implementing social network strategies for HIV testing within community-based prevention agencies [2]. In this manuscript, we describe the development of the Hep-Net intervention and its implementation and evaluation through a pilot randomized controlled trial (RCT). We present baseline data describing the participants enrolled and discuss challenges encountered disseminating the intervention using peer-based referrals.

Background

Epidemiologic studies suggest that HCV transmission is increasingly driven by injection drug use among young adults in rural and suburban settings. A cluster investigation in 6 contiguous rural counties in northern Wisconsin found that the number of HCV infections reported annually increased by more than 200% between the periods 2004-2008 and 2009-2010 [3]. Among individuals newly diagnosed with HCV in this outbreak, 94% reported a history of sharing needles or other drug preparation equipment. In this investigation and in similar outbreaks in Massachusetts [4], rural Indiana [5], and several Appalachian states [6], many young adults described a history of injecting prescription opioid medications for several years before transitioning to injecting heroin or methamphetamine. These sharp increases in HCV incidence concentrated in communities with traditionally poor access to prevention services highlight the need for evidence-based, targeted interventions to reduce HCV transmission and coordinate efforts to increase HCV testing and linkage to treatment for those who are infected [7].

Syringe exchange programs are a widely used strategy to reduce harm related to injection drug use. Numerous observational studies support the effectiveness of syringe programs for reducing behaviors leading to transmission of HIV and viral hepatitis and increasing entry into drug treatment programs [8-11]. Ensuring the availability of sterile syringes and other drug injection equipment, while a necessary component of disease prevention for people who inject drugs, is only one of several strategies that can be implemented through syringe exchange programs [12,13]. Other important components of risk reduction include linkage to addiction treatment, overdose prevention, and testing and linkage to care for HIV, viral hepatitis, and sexually transmitted infections. However, resource limitations pose challenges to consistently delivering multicomponent services that meet the diverse needs of people who inject drugs. Syringe exchange programs face resource limitations that are driven by social and political factors such as prohibitions on federal funding and local opposition. Many syringe exchange programs have insufficient resources to provide adequate syringe coverage or deliver a full package of preventative services to their clients [14]. Further, even when prevention services are available in the community and are of no cost to clients, many high-risk individuals still cannot or do not access syringe exchange programs due to myriad environmental and psychosocial barriers. As a result, many people who inject drugs are not regularly engaged in prevention services [15].

Novel Approaches

Computerized Interventions

Computer-based interventions deployed in syringe exchange programs or other community-based settings may represent a promising, low-cost strategy for delivering tailored health information that is specific to the needs of people who inject drugs. Studies examining computer-tailored interventions (CTIs) have shown positive behavior changes in a wide range of contexts, including alcohol reduction in college students, preconception care in women, and HIV prevention among juvenile offenders and drug users [16-19]. A meta-analysis of 88 CTIs showed a significant effect size for behavior change in smoking cessation, mammography, physical activity, and dietary practices, indicating CTIs have a clinically significant impact on rates of behavioral risk factors [20].

CTIs assess individual behavior, environmental barriers, and psychosocial determinants of positive behavior change. They then use data-driven decision guidelines to construct automatic, tailored feedback providing each individual with a personalized approach to risk reduction. CTIs are mobile, user-friendly, and brief. As such, CTIs may have an advantage in engaging transient, hard-to-reach populations in resource-constrained prevention settings such as syringe exchange programs.

Extending Prevention Services Through Social Networks

Strategies to increase engagement in prevention services must address environmental barriers such as geographic inaccessibility and psychosocial barriers related to individual motivation and behavioral skills. Social network-based strategies, which have been developed and implemented in many US cities to increase HIV testing, may facilitate dissemination of prevention services through both of these domains. In a demonstration project funded by the Centers for

Disease Control and Prevention (US Department of Health and Human Services) conducted in 7 US cities, 5.6% of clients recruited through peer referrals were HIV positive compared to a prevalence of approximately 1% who self-referred [21-23]. Programs to promote HIV testing have taken advantage of existing social networks and the meaningful influence of peers by enlisting high-risk clients to recruit, refer, or otherwise encourage their associates to participate in testing. Adaptation of this strategy to deliver HCV testing and prevention services to people who inject drugs was one of the factors motivating the development of this project.

Methods

Study Design

The Hep-Net Intervention: Overall Objectives

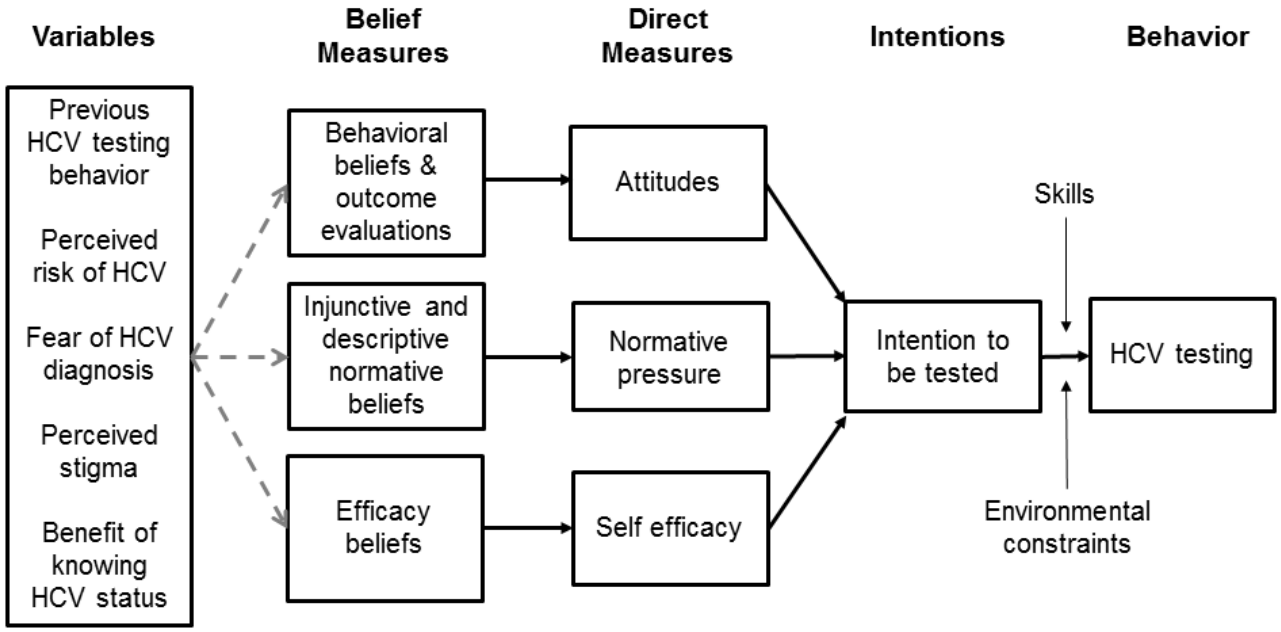
This project has two main objectives. First, it aims to determine whether a CTI is a feasible and acceptable approach to increasing readiness to engage in various health-promoting behaviors among people who inject drugs. It targets 4 different behavioral domains: (1) undergoing regular HCV screening, (2) using clean works for every injection, (3) taking steps to prevent opioid overdose, and (4) reducing and ultimately ceasing injection drug use. The second objective is to determine the feasibility of using social networks to expand delivery of computerized prevention interventions to hard-to-reach people who inject drugs.

Theoretical Frameworks

Hep-Net is grounded in behavior change theory and motivational interviewing techniques. The guiding behavioral theory is the integrative model of behavior change, which is schematized in Figure 1 [24,25]. The integrative model and its historical predecessors, the theory of planned behavior [26] and the theory of reasoned action [27], are supported by evidence across a wide array of health behaviors and populations [28-31]. The integrative model expands the scope of the theory of planned behavior and the theory of reasoned action by acknowledging the importance of skills, abilities, and environmental constraints as moderators of the relationship between behavioral intention and action [32].

Hep-Net is also guided by the transtheoretical model (ie, stages of change), which assumes that behavior change should be considered a continuum rather than a dichotomy [33,34]. Specifically, behavior change occurs through a series of stages in which individuals can move back and forth. In the precontemplation stage, individuals are not yet considering behavior change. In the contemplation stage, individuals may be considering change but have not yet taken steps toward behavioral change. Contemplation is followed by preparation, action, and maintenance. Although the stages are considered serial, one may skip particular stages (eg, planning/preparation) and, at any point in the continuum, one may digress to a previous stage of readiness (ie, from action to preparation). The Hep-Net system uses motivational interviewing techniques, which are founded on the transtheoretical model, to assess readiness for change with respect to safe injection practices, substance use reduction, and overdose prevention. The model also informed the types of specific feedback and risk reduction activities suggested to participants, as described below in the discussion of the risk reduction exercise.

Figure 1. The Integrative Model (adapted from Fishbein [24]).



Formative Research and Intervention Development

Using the integrative and transtheoretical models as the guiding frameworks, the system assesses participants' risks and protective behaviors; preferences for behavior change domain (eg, decreasing risky injection practices, reducing opioid use, overdose prevention); and attitudinal, normative, and efficacy beliefs. The system then tailors the content of the intervention to the individuals' stage of readiness for change, salient beliefs, and the chosen behavioral target (eg, safer injection practices).

To develop appropriate questionnaire items and health-promotion messages that address relevant beliefs held by the target population, we analyzed formative data collected through an anonymous cross-sectional survey of 553 syringe exchange clients [1]. Health promotion messages specifically tailored to individual stages of readiness to change were developed collaboratively by a research team of investigators with expertise in health communications, counseling psychology, and clinical medicine. Draft messages were discussed at in-person meetings and later pilot tested with several end-users who were current or previous clients of the syringe exchange program to elicit feedback to inform further refinement of messages.

The risk assessment survey and tailored behavioral computerized intervention were delivered using an Internet-based, customizable, interactive software tool developed in consultation with DatStat Inc (Seattle, WA). DatStat performed all necessary programming for creation of the intervention. The tool provided a patient-driven counseling experience similar to those used in other studies of high-risk populations [35,36]. The intervention simulated a motivational interview, asking contextually appropriate questions, and was intended to capture the essence of a patient clinical experience on a computer. In real time, the program synthesized patient responses about risks, knowledge, and beliefs; presented a list of risk factors to the participant based on those responses; and then guided the participant in developing an individualized risk reduction plan.

After developing and pilot testing the intervention with a small number of volunteers, we proceeded to recruitment and enrollment in the pilot RCT. The study protocol was reviewed and approved as a minimal risk study by the Health Sciences Institutional Review Board at the University of Wisconsin-Madison.

Study Population, Recruitment, Eligibility, and Screening

Enrollment in the study began in September 2014, and all baseline assessments were completed by April 2015. Participants in the study were either clients of an established, multisite syringe exchange program operating in southern Wisconsin or peers recruited from the social networks of these clients. Eligibility criteria included age 18 years or older, injection drug use in the past 30 days, and willingness to provide contact information for the 3-month follow-up. Pregnant women and people who did not speak English were excluded. As one goal of the study is to conduct outreach among high-risk populations who do not regularly use prevention services, we used social network–based referrals to recruit the majority of the study sample. Syringe exchange clients were informed about the study and screened for eligibility during a routine encounter at the syringe exchange program. Upon completion of the baseline visit, study participants received referral coupons and were encouraged to refer eligible peers. Coupons were marked with a unique code number used to track referral chains. Participants received US $10 in cash as compensation for time spent completing the baseline study encounter and an additional US $10 for each eligible peer they referred (up to 5) who enrolled in the study.

Baseline Study Assessment

At the initial study encounter, participants were encouraged to get a rapid HCV antibody test unless they had had a positive HCV antibody test in the past or had gotten a rapid HCV test within the last 3 months. Receiving an HCV test was not a requirement for participation in the study; it is a standard service offered to all syringe exchange clients. The computerized survey was designed to last 20 to 30 minutes. For those consenting to HCV testing, the baseline assessment was administered after participants provided a fingerstick blood specimen for the rapid HCV test, allowing them time to complete most of the baseline assessment while awaiting the test results. The complete baseline questionnaire is reproduced in Multimedia Appendix 1.

Question items were developed based on the integrative model to evaluate attitudes, norms, and self-efficacy beliefs relevant to each of the targeted health behaviors. Table 1 displays sample question items assessing the relevant constructs in the integrative model for the behavior of HCV testing. Response options to each of these questions were a 5-point Likert scale ranging from "strongly disagree" to "strongly agree." Participants rated their readiness to make changes toward each of 4 behavioral goals: (1) "I will cut down on my drug use or quit using drugs completely," (2) "I will use clean needles, cottons, and cookers every time I inject drugs," (3) "I will get tested for hepatitis C every 6 months for as long as I'm using," and (4) "I will get trained to give naloxone (or Narcan) in case someone I am with has an overdose."

Table 1. Examples of questions based on the integrative model.

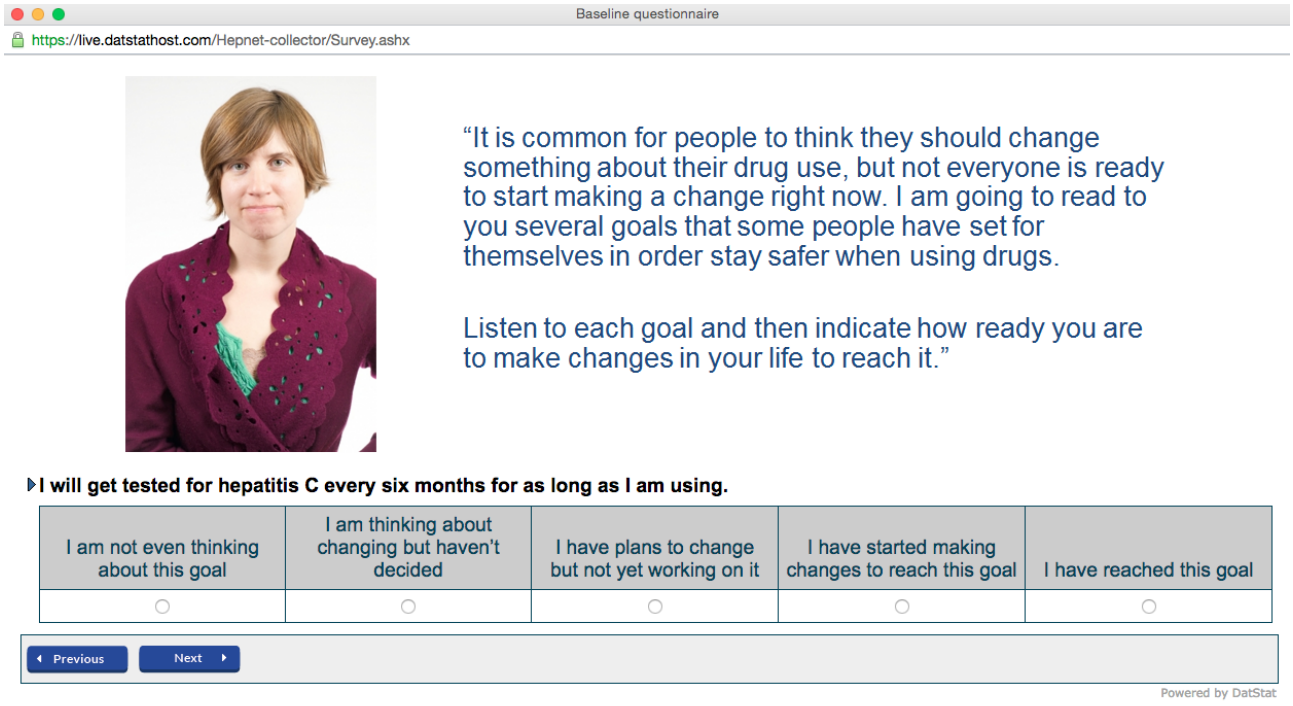
Survey question	Integrated model domain
"I am confident that if I really wanted to, I could get tested for hepatitis C every six months, for as long as I am shooting drugs."	Self-efficacy
"Most people who are important to me think I should get tested for hepatitis C."	Injunctive normative beliefs
"Most people who are similar to me have been tested for hepatitis C."	Descriptive normative beliefs
"Getting tested for hepatitis C is important to me."	Attitudes

Using the transtheoretical model, we assessed readiness to adopt specific healthy behaviors using the visual analog scale shown in Figure 2. Each behavior was characterized as a health-related goal that a person who injects drugs may have, and respondents were asked to characterize their readiness to adopt the behavior by selecting a statement on the spectrum of "I am not even thinking about this goal" (precontemplation stage) to "I have reached this goal" (maintenance stage). For the intervention

group, the stage of change reported by respondents during the baseline session was used in the algorithms to determine what tailored content would be displayed in the subsequent risk reduction intervention.

The baseline questionnaire included basic demographic and locator information, including multiple means of electronic communication (eg, text messaging, email, Facebook) to facilitate coordination of follow-up. Additional sections included a risk behavior questionnaire assessing addiction severity, overdose risk, and injection-related and sexual behaviors associated with transmission of HCV. To maximize accurate disclosure of high-risk and sensitive behaviors, both the baseline questionnaire and the tailored intervention used audio computer-assisted self-interview. As an enhancement designed to better simulate a motivational interviewing session, photographs of a model portraying a counselor were embedded in the survey program to accompany the audio-recorded instructions, survey questions, and, if applicable, tailored feedback messages. At the beginning of the session, participants selected 1 of 3 female avatars that appeared to have varying racial/ethnic backgrounds. The avatar selected by the participant would be displayed throughout the baseline session and the subsequent follow-up assessment 3 months later.

Figure 2. Assessment of readiness to change based on the Transtheoretical Model.



Randomization

After completion of all required sections of the baseline survey, participants were randomly assigned to receive the risk reduction intervention or be in the control group, in which the computer session terminated after completion of the survey. Both the intervention and control groups received individualized prevention counseling per standard of care at the syringe exchange programs. Challenges related to data synchronization across multiple sites and the need for offline data collection in rural communities made stratified, block randomization infeasible. Therefore, simple randomization was used to place participants to either group using the survey software during the baseline session.

Intervention Content

Overview

Participants assigned to receive the risk reduction intervention were presented with a series of screens featuring text and audio content summarizing the participant's risk behaviors and delivering health-promotion messages. Components of the tailored intervention consisted of an overall risk synthesis, selection of behavioral goal, and individualized risk reduction exercise and assessment of self-efficacy related to risk reduction plan. An example of the series of content screens displayed for a participant in the precontemplation stage with respect to overdose prevention is reproduced in Multimedia Appendix 2.

Risk Synthesis

The risk synthesis was introduced with 1 screen displaying positively framed feedback messages emphasizing behaviors reported by the participant that can reduce risk of HCV transmission and/or opioid overdose. A subsequent screen displayed tailored feedback regarding specific behaviors associated with increased risk reported by the participant.

Selection of Behavioral Goal

In the second portion of the intervention, participants chose a behavioral goal they would like to work on over the next 3 months (HCV testing, Narcan training, use of clean needles or works, or reducing or suspending drug use) and selected action steps tailored to the participants' assessed stage of change within that one risk category. The relevant stage of change was determined by responses to questions addressing the risk reduction goals.

After selecting a behavioral risk reduction goal, participants received a message introducing the activity that was tailored to their self-reported stage of readiness to change, relevant to that particular risk category. Participants were then guided through a series of screens which gave feedback and educational content (eg, the opportunity to view a brief video) tailored to the types of risk behaviors and stage of readiness to change. Messages were designed to encourage movement along the stages of change in the direction of action/maintenance.

Individualized Risk Reduction Exercise

The final intervention component was an interactive activity in which participants were asked to create a risk reduction plan. Participants were presented with a list of 10 to 12 possible suggested action steps related to the risk reduction goal they selected. The content of these lists was informed by the transtheoretical model, encouraging incremental change through concrete actions that could be taken. Participants were asked to select 3 to 5 of the steps they felt they would be able to do in the ensuing 3 months. After participants selected the steps, their individualized risk reduction plan was read back to them. They were asked to confirm their selected steps and given an opportunity to go back and change their choices. Finally, participants were asked to answer a series of questions assessing confidence in their ability to complete their plan.

Follow-Up Assessments and Outcome Measures

Upon completion of the baseline session, participants were reminded of the need to return for a follow-up assessment 3 months after the baseline session. The second study visit uses an Internet-based survey designed to assess the same behavioral and attitudinal domains captured through the baseline questionnaire. This allows us to evaluate any temporal changes in the frequency of substance use, injection risk behaviors, overdose risk, and HCV testing. The follow-up assessment also evaluates self-reported readiness to change, attitudes, and perceived norms, providing an opportunity to detect whether the tailored intervention may influence outcomes through these intermediate variables.

In addition to repeating the assessment of baseline variables for longitudinal analysis, the follow-up questionnaire captures information on participant perceptions about whether they met any of the health goals discussed in the baseline session. For participants determined to be HCV-infected at baseline, the survey assesses whether they received any follow-up testing or medical care for HCV since receiving their test result. Finally, it evaluates usability and acceptability of the intervention through a series of questions delivered to participants who were randomized to the active study arm. The additional questionnaire items used during the follow-up assessment are presented in Multimedia Appendix 3.

There are multiple behavioral outcomes of interest in this project. Accurate assessment of drug use behaviors may be limited because it relies on participant self-report and may be biased due to losses to follow-up. HCV testing behavior may be captured with greater validity because it will be ascertained by searching HCV testing data collected and reported by all agencies receiving funding from the Wisconsin Division of Public Health. For participants who have received a reactive HCV screening test result, we will determine whether any follow-up testing was performed, including confirmatory HCV RNA or HCV genotype tests, which would indicated that the participant was linked to evaluation and/or treatment of HCV.

Sample Size and Power

Demonstrating efficacy of the intervention through detection of a statistically significant effect size was not a primary goal of the project. However, we considered it plausible that the intervention might significantly influence participants' decisions to receive HCV testing even after a single session. While planning the pilot trial, we calculated a target sample size that would be sufficient to detect what we considered to be a meaningful difference in the proportion of participants who undergo HCV testing within 12 months after enrollment. Assuming that approximately 10% of participants would be known to have HCV infection at enrollment and accounting for expected losses to follow-up, we estimated that a sample size of 408 would provide 90% power to detect a difference of 0.15 in the proportion of participants who voluntarily returned for an HCV test within a year of enrollment.

We believed that achieving a target sample size of 408 was feasible and justified based on data from the pilot survey conducted in 2012 reporting that 69.4% of syringe exchange clients had an HCV test in the prior 12 months and 14.9% of clients had ever had a positive HCV test [1]. Because this intervention was targeting people who inject drugs but may not be regular users of the syringe exchange program, we anticipated that the number reporting prior HCV testing would be lower. We believed an effect size of this magnitude was reasonable based on prior meta-analyses of tailored communication interventions, which provide support for moderate mean intervention effect sizes [20] across a variety of health behaviors, including addiction-related behaviors (eg, smoking) and HIV risk-related behaviors [37]. Notably, findings suggested effect sizes increased with the number of behaviors intervened upon, with mean effect size of g=.24 (95% CI 0.18-0.31) for interventions focused on 3 behaviors [20].

Data Analysis

Descriptive statistics reported here were calculated for baseline variables using SAS 9.3 (SAS Institute Inc). Differences in participant characteristics were assessed using a Wilcoxon rank-based general linear model approach for continuous variables and Pearson chi-square or Fisher exact tests for differences in categorical variables.

To evaluate the effectiveness of the risk reduction intervention, we will test within-group changes from baseline to follow-up (eg, change in frequency of needle-sharing) using the matched-pair Wilcoxon rank-sum test. Similarly, binary outcome changes will be examined using either the McNemar test or Fisher exact test.

To assess the independent role of the intervention in improving screening for HCV, we will use a logistic regression model on the binary outcome of having an HCV test within the 12-month period postintervention. Again, a Wilcoxon general linear model framework will be used for analysis of Likert scale outcome

measures. Indicators pertaining to individual level outcomes within intervention and control groups will be measured among the same individuals at baseline and follow-up.

To avoid potential sampling bias toward individuals with larger networks, we will adjust regression models using inverse probability weights based on individual recruitment network sizes [33]. To account for correlation between recruiter and recruited, we will create a variable indicating who the recruiter of each subject was and use it as a cluster variable using generalized estimating equations. An exchangeable correlation structure within each cluster will be assumed (ie, correlation between any 2 subjects recruited by the same recruiter will be assumed to be the same). For all tests, a 2-sided *P* value of less than .05 is considered statistically significant.

Results

Between September 2014 and April 2015, 235 people completed the baseline survey. Baseline descriptive characteristics of the sample are displayed in Table 2. The racial/ethnic breakdown was reflective of the general population of the region, with non-Hispanic whites comprising 60.3% (129/232) of the sample and those self-described as black or African American comprising 28.1% (66/235). The median age was 35 years (range 18-63 years). While most participants who provided a valid address (131/219, 59.8%) lived in the city of Milwaukee, about one-third (70/219, 29.0%) of participants resided in a municipality with a population less than 50,000 residents, including 17.8% (39/219) who lived in a city with a population less than 5,000. Comparison of the control and intervention groups with respect to baseline characteristics demonstrated no statistically significant differences, assuming a 2-sided alpha of .05.

Table 2. Baseline characteristics by intervention group (N=235).

Characteristics	Category	Control (N=126)	Intervention (N=109)
Age, years, median (IQR)		35 (28-46)	33 (27-44)
Gender, n (%)	Male	92 (73.0)	89 (81.7)
	Female	34 (27.0)	20 (18.3)
Race, n (%)	White	79 (63.0)	59 (54.1)
	Black	32 (25.0)	34 (31.2)
	Other or multiple	15 (12.0)	16 (14.7)
Ethnicity, n (%)	Non-Hispanic/Latino	112 (90.3)	102 (94.4)
	Hispanic/Latino	12 (9.7)	6 (5.6)
Highest education level, n (%)	Less than high school	17 (13.5)	7 (6.4)
	HS diploma or GED	61 (48.4)	51 (46.8)
	Some college or vocational school	45 (35.7)	47 (43.1)
	College degree	3 (2.4)	4 (3.7)
Currently employed, n (%)	No	92 (73.0)	69 (64.0)
	Yes	34 (27.0)	39 (36.0)
Legal income in last year, n (%)	None	31 (26.0)	30 (27.5)
	US $1-11,500	62 (51.0)	49 (45.0)
	More than US $11,500	28 (23.0)	30 (27.5)
Homeless during the past year, n (%)	No	58 (46.0)	52 (48.0)
	Yes	68 (54.0)	56 (52.0)
Incarcerated during the past year, n (%)	No	82 (66.0)	63 (59.0)
	Yes	42 (34.0)	44 (41.0)
Has health insurance, n (%)	No	20 (16.0)	11 (10.0)
	Yes	105 (84.0)	98 (90.0)
Has primary care provider, n (%)	No	53 (42.4)	39 (36.0)
	Yes	72 (57.6)	69 (64.0)

Of the 235 participants who completed the baseline assessment, 80 (34.0%) agreed to receive a rapid HCV test, and 14 (17.5%) tests were reactive. The most common reasons given for declining the test were the participant had been previously tested

(36/155, 23.2%), did not want to or did not feel ready to be tested (22/155, 14.2%), already knew he or she was HCV-positive (17/155, 11.0%), and did not have enough time (13/155, 8.0%).

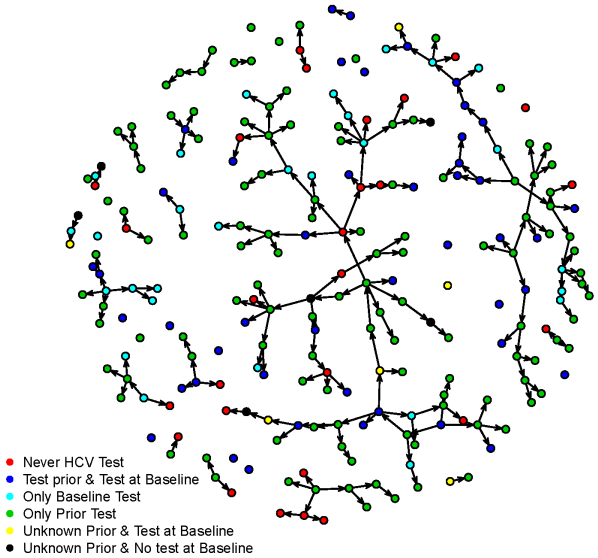
Drug use characteristics of the baseline sample are displayed in Table 3. Heroin was the drug most frequently injected by participants and nearly half (105/235, 44.7%) reported that they inject on a daily basis. Most participants (161/234, 68.8%) reported they had shared syringes, cotton filters, or cookers with other people while injecting drugs in the past. Of those reporting sharing injection equipment in the past, 44.1% (71/161) had shared syringes, cottons, or cookers during the past 3 months, and 30.4% (49/161) reported they had never been tested for HCV.

Table 3. Drug use characteristics by intervention group (N=235).

Characteristics	Category	Control (N=126) n (%)	Intervention (N=109) n (%)
Drugs injected in past 30 days	Heroin	111 (88.1)	92 (84.4)
	Prescription opioids	26 (20.6)	21 (19.3)
	Cocaine	36 (28.6)	35 (32.1)
	Methamphetamine	1 (0.8)	6 (5.5)
Frequency of drug injecting in past 30 days	Less than daily	67 (53.2)	63 (57.8)
	Every day	59 (46.8)	46 (42.2)
Has shared needles, cottons, or cookers	No	37 (29.6)	36 (33.0)
	Yes	88 (70.4)	73 (67.0)
Has had an opioid overdose	No	75 (60.5)	70 (64.2)
	Yes	49 (39.5)	39 (35.8)

Using social networks to recruit participants allowed us to reach a population that may have otherwise not been reached. In the first phase of the study, prevention staff recruited 40 individuals to participate who were existing clients of the syringe exchange program. These participants referred 195 peers who were determined to be eligible and were enrolled in the study. As shown in Figure 3, linking participants via referral chains allows visualization of 2 large networks and several smaller ones.

Figure 3. Network diagram of peer referral chains by hepatitis C virus testing history.



Discussion

Main Findings

The overarching goal of this pilot RCT was to determine if a computerized, tailored intervention was feasible and acceptable to implement in an syringe exchange program. Though its potential impact in increasing HCV testing, overdose prevention, and use of clean syringes and works will be reviewed after all follow-up assessments and HCV testing follow-up data are collected, the data collected to date demonstrate that the approach is feasible overall. There was a high level of willingness to participate, and many participants referred peers, indicating acceptability of this type of intervention.

Unanticipated Challenges

Throughout the initial period of enrollment and completion of baseline assessments, the study staff encountered several unanticipated challenges. Scheduling appointments for enrollment and completion of study assessments proved to be difficult; participants preferred to drop in at a time that was convenient for them. Often, individuals forgot about their appointments, did not have transportation to make it to their scheduled appointment, and could not be reminded via phone or text because they did not own a cell phone or have minutes. Both study sites amended their procedures and became accessible for drop-in appointments to accommodate the difficulties faced by people who inject drugs in keeping scheduled appointments.

The Hep-Net study was designed as a pragmatic intervention trial rather than a highly controlled clinical trial, which would have required more resources and been less generalizable to other settings. Prevention staff had numerous responsibilities that superseded the tasks they were asked to complete for the research project, such as obtaining informed consent and administering the computerized survey. Balancing these competing demands required communication and an effective partnership among the research team, the existing prevention staff, and administrators of the syringe exchange program. Within 2 months of enrolling the first participants, study site staff began to identify the best time periods for survey visits and how to best fit the intervention into routine prevention services. When the study team and prevention staff determined how to best achieve balance between research and service activities, it became obvious that the most appropriate rate of accruing new subjects would not allow recruitment of a sample as large as originally planned. The target sample size was modified after several months for this reason to the more realistic goal of 120 subjects per group.

Computer literacy was another unanticipated challenge experienced by study staff. Many participants had little background using computers and struggled to understand how to answer questions and advance the program. Although site staff made themselves available for computer questions and aided participants in computer fluidity, several participants took over an hour to complete the survey rather than the anticipated half hour.

Finally, Internet connectivity and a private space to screen and deliver the survey were difficult barriers to overcome. Neither syringe exchange program had readily available WiFi, so Ethernet accessibility was imperative in collecting and synchronizing survey data. Private spaces with Ethernet accessibility were difficult to keep consistently vacant of other prevention services, which made data collection more difficult. Furthermore, a substantial number of study assessments were completed via mobile syringe exchange outreach where no Internet connectivity was available. The remote data collection feature in the DatStat software package was used for these surveys, which added a layer of complexity causing some frustration among participants and staff.

The main limitation of this study is its lack of generalizability to other cities and states and other local epidemics. While Hep-Net may be feasibly used in the context of Wisconsin's opioid epidemic, it may not apply to other geographical areas targeting the same population. This CTI used existing preliminary data from a pilot study of over 500 Wisconsinites to tailor the intervention content to a specific population. Additionally, our sample was subject to selection bias because individuals using a syringe exchange program tend to be a healthier, higher-functioning subset of people who inject drugs. Although the computerized approach to data collection was designed for complete anonymity to reduce social desirability bias, participants may have answered questions to please researchers.

A major strength of this study is the use of well-established syringe exchange programs as home to Hep-Net. The community-based prevention specialists who implemented this project are highly regarded among community members and have built trust over years of service. Without their involvement, acceptability of the intervention would likely have been much lower.

Conclusion

If effective, Hep-Net has the possibility to facilitate a more comprehensive approach to prevention and linkage to care within syringe exchanges and other community-based programs. Syringe exchange programs, while shown to be effective, are already understaffed and lack resources. Hep-Net's role is to fill in the gaps presented by agency challenges to provide behavioral care to a subset of a community that is substantially underserved. To disseminate the intervention, busy syringe exchange programs could present Hep-Net to clients or use the Internet to reach rural or immobile populations. We hope the results will lead to implementation of a CTI in community-based settings. If the study hypotheses are confirmed, the proposed solution can be tailored to specific cities and states and disseminated to reduce the impact of hepatitis C among people who inject drugs.

Conflicts of Interest

None declared.

Multimedia Appendix 1

Baseline questionnaire.

[PDF File (Adobe PDF File), 193KB - resprot_v5i1e15_app1.pdf]

Multimedia Appendix 2

Sample content screens from Hep-Net intervention.

[PDF File (Adobe PDF File), 769KB - resprot_v5i1e15_app2.pdf]

Multimedia Appendix 3

Follow-up assessment.

[PDF File (Adobe PDF File), 19KB - resprot_v5i1e15_app3.pdf]

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Abbreviations

CTI: computer-tailored interventions
HCV: hepatitis C virus
RCT: randomized controlled trial

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Protocol

Increasing Access to Mental Health Care With Breathe, an Internet-Based Program for Anxious Adolescents: Study Protocol for a Pilot Randomized Controlled Trial

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Abstract

Background: There is a demand to make first-line treatments, including cognitive behavioural therapy (CBT) for adolescent anxiety disorders, more widely available. Internet-based CBT is proposed to circumvent access and availability barriers and reduce health care system costs. Recent reviews suggest more evidence is needed to establish the treatment effects of Internet-based CBT in children and adolescents and to determine related economic impacts.

Objective: This pilot trial aims to collect the necessary data to inform the planning of a full-scale RCT to test the effectiveness of the Internet-based CBT program Breathe (Being Real, Easing Anxiety: Tools Helping Electronically).

Methods: We are conducting a 27-month, 2-arm parallel-group, pilot randomized controlled trial (RCT). Outcomes will inform the planning of a full-scale RCT aimed to test the effectiveness of Internet-based CBT with a population of adolescents with moderate to mild anxiety problems. In the pilot RCT we will: (1) define a minimal clinically important difference (MCID) for the primary outcome measure (total anxiety score using the Multidimensional Anxiety Scale for Children); (2) determine a sample size for the full-scale RCT; (3) estimate recruitment and retention rates; (4) measure intervention acceptability to inform critical intervention changes; (5) determine the use of co-interventions; and (6) conduct a cost-consequence analysis to inform a cost-effectiveness analysis in the full-scale RCT. Adolescents aged 13-17 years seeking care for an anxiety complaint from a participating emergency department, mobile or school-based crisis team, or primary care clinic are being screened for interest and eligibility. Enrolled adolescents are being randomly allocated to either 8 weeks of Internet-based CBT with limited telephone and e-mail support, or a control group with access to a static webpage listing anxiety resources. Adolescents are randomly assigned using a computer generated allocation sequence. Data are being collected at baseline, treatment completion, and at a 3-month follow-up.

Results: Currently, adolescents are being enrolled in the study. Enrolment is taking place between March 2014 and February 2016; data collection will conclude May 2016. We expect that analysis and results will be available by August 2016.

Conclusions: In many communities, the resources available for front-line anxiety treatment are outweighed by the need for care. This pilot RCT is an essential step to designing a robust RCT to evaluate the effectiveness of an Internet-based CBT program for adolescents with moderate to mild anxiety problems.

Trial Registration: Clinicaltrials.gov NCT02059226; http://clinicaltrials.gov/ct2/show/NCT02059226 (Archived by WebCite at http://www.webcitation.org/6epF8v7k4)

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KEYWORDS

anxiety; etherapy; cognitive behavioral therapy; adolescents; mental health; Internet; intervention; pilot; randomized controlled trial

Introduction

Anxiety disorders are the most common mental disorders diagnosed in adolescence, with prevalence before age 18 reported as greater than 30% [1,2]. The median age of onset is 11 years, making these disorders some of the earliest to develop [3]. Early onset disorders often follow a chronic course [4] and can significantly interfere with interpersonal relationships, academic performance and daily functioning [5-7]. Anxiety disorders range in severity and specific symptomology, but the more frequently diagnosed disorders in adolescents include social anxiety disorder (social phobia) [8-10], generalized anxiety disorder [8,11,12], and specific phobia [8,12]. The most prevalent is social anxiety disorder, with studies showing a two-fold increase in the prevalence of this disorder during teenage years [13,14]. The high prevalence of social anxiety disorder in adolescence can be understood in the context of developmental and environmental transitions (eg, puberty, dating, new schools, peer experiences) [9]. Undetected or undertreated, these disorders present significant future risk for adult anxiety disorders, educational underachievement, suicidality, depression, substance abuse and future hospitalization [5-7,15-19].

Cognitive behavioral therapy (CBT) as a first-line treatment for anxiety disorders in children and adolescents is well established [20,21,22] and supported by meta-analytic work [23,24,25]. CBT combines systematic exposure to feared situations with skills training and the learning of activities to help replace anxious thoughts about feared situations with more adaptive thoughts [20,21]. These treatment elements are based on the premise that repeated exposure to feared situations results in desensitization, reducing anxiety and avoidant behaviors, and improving functioning. Relaxation strategies, such as imagery and deep breathing, are included in skills training to provide ways to manage discomfort during repeated exposures [21].

Trained mental health professionals have traditionally delivered CBT, but the structured and sequential nature of CBT translates well to computer-based delivery. Within this approach, the use of the Internet as a delivery mode provides researchers with unobtrusive ways to capture data and examine access patterns, compliance, and usability issues [26]. Internet-based delivery also circumvents multiple barriers to receiving in-person CBT including: social stigma, direct and incidental costs (eg, time out of school), lack of trained deliverers, and inconvenient service times and locations [27]. This delivery method may also increase the number of young people seeking professional help. A published paper in 2002 reported that over a one-year timeframe, only 8.3% of young people with recent onset anxiety and 21.4% with chronic anxiety had sought professional help [28]. While Internet-based CBT is considered lower-intensity than in-person treatment, the majority of adolescents with anxiety disorders experience mild to moderate distress and impairment, making treatment availability paramount. Thus, Internet-based CBT is proposed to reduce demand for therapy delivered in-person, introduce cost savings, and increase timely access to recommended treatment [29].

Three recent systematic reviews of the literature have shown that self-led, computer-based and computer-assisted CBT, in combination with in-person or therapist-guided care, are viable and effective treatment approaches for children and adolescents [30,31,32]. Studies of treating adolescents using Internet-based CBT are limited to two [33,34], but provide important evidence and methodological considerations for other treatment studies. In a study targeting social fears (mainly centered on public speaking) in high school students who met the diagnostic criteria for social anxiety disorders, Tillfors et al. found that significant reductions in anxiety symptoms and improvements to mood were achieved post-intervention and maintained at one-year follow-up [34]. These treatment effects were noted as comparable to those achieved through traditional face-to-face CBT. Focusing on the treatment of adolescents who had a "markedly disturbing/disabling" anxiety disorder (predominantly generalized anxiety disorder and social phobia), Spence et al. also found that Internet-based CBT with minimal therapist support was equally efficacious as face-to-face CBT delivery in reducing anxiety symptoms post-intervention and up to one-year post-treatment [33]. The parents of participating adolescents also took part in this online program. In both studies, methods to improve treatment compliance, and the impact of compliance on treatment outcome, were noted as issues that required further investigation. Tillfors et al. reported that, of nine online modules available to complete within a 9-week period, the average number of completed modules was 2.9 [34]. Spence et al. noted that on average, adolescents and parents completed 7.5/10 and 4.48/5 sessions, respectively [33].

While the reported treatment effects of Internet-based CBT for treating adolescent anxiety are promising [33,34], published systematic reviews highlight important methodological issues that future studies must consider, in order to mature the evidence base [30,31,32]. Two reviews reported that the overall quality of the outcomes reported by studies of computerized CBT, including Internet-based CBT for adolescents, was *low* to *very low* [30,32]. Key limitations of the evidence base that were identified included: small sample sizes, intervention and comparator (waitlist or active control) differences between studies, and inconclusive treatment effects when anxiety was self-rated (versus clinician rated) [30,31,32]. Many of these limitations were also noted in the Tillfors and Spence studies

[33,34]. Reviews to date have recommended that higher quality evidence is needed to firmly establish the treatment effects of computerized CBT, and that the study of associated economic impacts is needed [30,31,32].

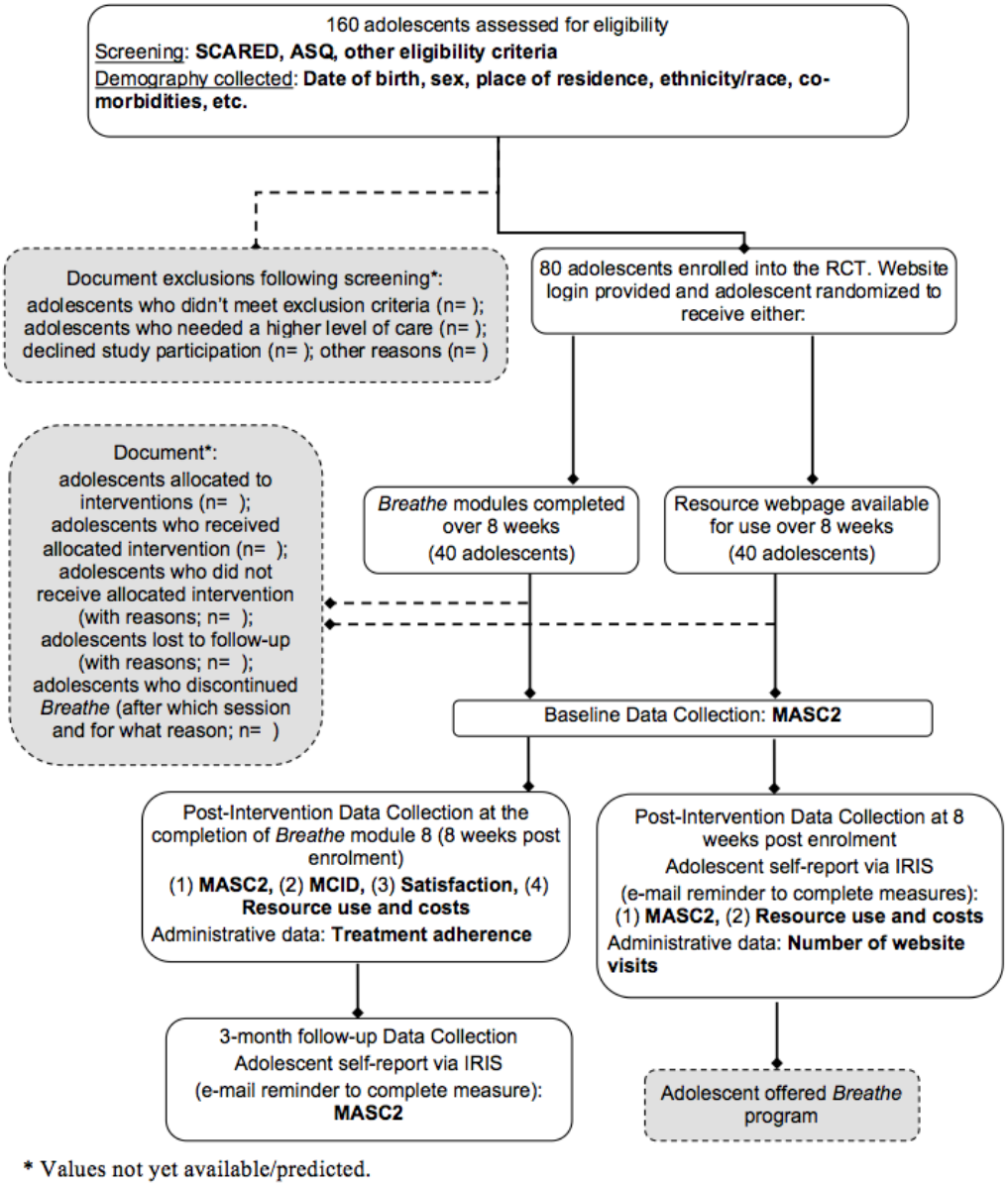
We designed a pilot randomized controlled trial (RCT) that addresses several of the limitations highlighted in the literature to date. This pilot trial aims to collect the necessary data to inform the planning of a full-scale RCT to test the effectiveness of the Internet-based CBT program *Breathe* (Being Real, Easing Anxiety: Tools Helping Electronically). In the pilot RCT we will (1) define a minimal clinically important difference (MCID), as defined by adolescents, for the primary outcome measure, (2) determine a sample size for a full-scale RCT, (3) estimate recruitment and retention rates to determine the number of study sites needed, (4) measure intervention acceptability to inform critical intervention changes, (5) determine the use of co-interventions, and (6) conduct a cost-consequence analysis to inform a cost-effectiveness analysis for a full-scale RCT.

Methods

Study Design

We are conducting a 27-month, 2-arm parallel-group, pilot RCT. Eighty adolescents will be randomly assigned to either 8 weeks of Internet-based CBT with telephone and e-mail support (the *Breathe* program), or a control (minimal intervention) group with 8 weeks of access to a static webpage with general anxiety resources and information. Study enrolment is taking place from March 2014 to February 2016; data collection will conclude May 2016. Details of the study design are illustrated in Figure 1. The study has been approved by host institutions' Research Ethics Boards.

Figure 1. Trial flow diagram.



Participants

Participants are adolescents from three Canadian provinces (Alberta, Ontario, and Nova Scotia) seeking care from a pediatric emergency department, mobile or school-based mental health team, or primary care setting that is participating as a study recruitment site. Study inclusion criteria are: (1) ages 13 to 17 years, (2) self-reported mild to moderate anxiety, (3) ability to read and write English, (4) regular access to a telephone and a computer system with high speed Internet service, and (5) ability to use a computer to interact with web material. Adolescents aged 13 to 14 years must also have a consenting parent.

Adolescents are screened for study inclusion criteria via a secure online recruitment process or telephone-based recruitment phone call. Demography is collected during the screening process to characterize adolescents who meet and do not meet clinical eligibility. Screening criteria (1), (3), (4), and (5) are assessed through yes/no answers. We are using the Screen for Child Anxiety Related Emotional Disorders (SCARED) [35] to screen for mild-to-moderate anxiety symptoms (eligibility criteria #2). The SCARED is a 41-item self-report screen for symptoms of panic disorder, social phobia and anxiety disorder, and general anxiety disorder in clinical and community adolescent samples, based on DSM-IV criteria [36,37,38]. Adolescents can report any experience of anxiety (ie, responds to any of the 41 items of anxiety symptoms/experiences as being "Somewhat/Sometimes True" or "Very/Often True") to be considered eligible for study participation.

We are also screening for risk of suicide using the Ask Suicide-Screening Questions (ASQ) [39]. The ASQ is a brief 4-item instrument originally validated for use in emergency departments with high sensitivity for identifying the risk of suicide [39]. A "yes" response to questions 1 ("In the past few weeks, have you wished you were dead?"), 2 ("In the past few weeks, have you felt that you or your family would be better off if you were dead?") or 4 ("Have you ever tried to kill yourself?") on the ASQ involves a safety-follow up phone call from a research team member to evaluate intent/severity/immediacy of risk before deciding on adolescent eligibility. Adolescents who respond "yes" to question 3 on the ASQ ("In the past week, have you been having thoughts about killing yourself?") are excluded from the study and receive brief telephone based support from a research team member to evaluate intent/severity/immediacy of risk and encourage the adolescent to seek further mental health care/crisis care.

Procedure and Randomization

Potentially eligible participants (and their parents) can receive oral, written, and/or online information about the study. In the Canadian provinces involved in this study, there is no specified age of consent. A minor's ability to consent or assent is decided after assessing capacity to consent. In this study, 15 to 17 year-olds will be allowed to consent to the study on their own behalf. We will require parental consent for adolescents aged 13 to 14 years. This decision was not based on any age of consent law; we want to acknowledge the role that parents have in their younger adolescent's activities and their ability to provide perspective on their child's potential participation in the study. Requiring parental consent will provide us with a formal mechanism to engage with parents before study enrolment. We will seek assent from adolescents aged 13 to 14 years even if they were assessed as being able to consent.

Randomization takes place after informed consent/assent is obtained. Adolescents are randomly assigned using a computer-generated allocation sequence. Participants are randomly allocated to one of two treatment conditions and receive this allocation information via e-mail from a graduate student trainee affiliated with the project. The e-mail also provides login/website information to the allocated intervention. Given the study objectives, the pilot was designed as open-label.

Measures

Primary Outcome Measure

Change in anxiety symptoms in adolescents will be measured using the total score from the Multidimensional Anxiety Scale for Children (MASC2; 50 items). The MASC2 is one of the most widely used self-report measures in clinical trials in adolescents with anxiety disorders. It assesses physical symptoms, social anxiety, harm avoidance, separation/panic, and total anxiety, and has excellent 3-month test-retest reliability [40] and excellent validity [41,42]. Adolescent self-report is desirable because of the internalizing nature of anxiety [43]. The MASC2 is administered online via Intelligent Research Intervention Software (IRIS) to adolescents in the treatment group at three data collection time points: baseline (pre-treatment), 8 weeks/end of 8th*Breathe* module (post-treatment) and 3 months post-treatment (follow-up). Adolescents in the control group complete the MASC2 at baseline and 8 weeks after study enrolment via IRIS. Table 1 outlines screening and outcome measurement time points.

Table 1. Outcome measurement schedule.

	Study time points					
	Study eligibility screening	Baseline	8 weeks (postintervention)	20 weeks (3-month follow-up)	End of study enrolment period (22 months from trial start)	End of data collection period (27 months from trial start)
Demography	X					
SCARED	X					
ASQ	X					
Co-intervention use/costs			X			
MASC2		X	X	X		
MCID			X			
Treatment satisfaction			X			
Treatment adherence			X			
Number of adolescents screened					X	
Number of adolescents enrolled					X	
Number of adolescents retained at 3-month follow-up						X

Secondary Outcome Measures

All secondary outcome data are collected via self-report using IRIS. The MCID will be determined for the primary outcome measure following module 8 completion (post-intervention). Adolescents allocated to the treatment condition are asked to indicate the minimum change in anxiety for which they would consider treatment participation. We will use the MCID to guide interpretation and application of outcome change in a full-scale RCT. To estimate the MCID, we will use adolescents' global ratings of change on a 10-point Likert scale (-5 to +5), a commonly used anchor [44,45].

Satisfaction will be measured in adolescents allocated to the treatment condition at the end of module 8 (post-intervention) to infer intervention acceptability. A 12-item instrument developed by the research team will measure satisfaction based on the adolescent's report of the program's engagement and sense of safety/privacy, expectations and usefulness, communication, and technical (intervention) management. For 10 statements, a 5-point Likert response format ranging from strongly agree (score 5) to strongly disagree (score 1) will be used. Scores range from 10 to 50 with scores of 40 or greater indicating higher acceptability. Two open-ended questions to inform the larger trial will allow adolescents to identify challenges or barriers that were faced in taking part in the trial, and the extent to which adolescents used the skills learned.

Treatment adherence will be measured at 8 weeks to further evaluate intervention acceptability. Adherence is measured by documenting the number of treatment modules and homework tasks completed. We will also record the number of tailored modules completed by each participant (treatment arm) and site visits (control arm). These data are being collected through IRIS.

Health care resource use will be detailed for a preliminary cost-consequence analysis [46,47]. The following are being reviewed: (1) software development and maintenance costs, (2) training and personnel costs for the treatment arm, (3) health care utilization data (eg, visits to the emergency department, hospital admission), and (4) other costs reported by adolescent (ie, time off from work/school). Adolescents will be asked at the post-intervention assessment to report on the use of co-interventions during the study period (eg, emergency department visits, other treatments, medication) and reasons for this use (eg, unmet need, medication prescribed before the study).

Sample Size

We have been conservative in estimating our ability to enroll and retain participants, by assuming that 50% will refuse to participate and 50% of enrolled adolescents will drop out. A total of 160 adolescents are being invited to participate with the expectation that we will enroll 80 adolescents (40 assigned to each study arm) and retain 40 adolescents (20 per arm) by module 8 completion (post-intervention). In keeping with the standard goals of a pilot RCT, a sample size of 40 adolescents at post-intervention (20 per arm) will provide sufficient data on the primary outcome to calculate a sample size based on 90% power for a full-scale RCT [48,49].

Treatment Condition

Essential Clinical Components

Consistent with published CBT treatment recommendations [20,21,22], the *Breathe* program consists of 8 core CBT treatment modules (8 weeks of treatment; Figure 2) that involve (1) multi-media based education about anxiety problems and approaches to overcoming anxiety (eg, reviewing why exposure exercises are important), (2) self-assessment activities to

determine level of treatment and safety needs (Figure 3), (3) activities to teach adolescents about anxiety sensitivity, how to identify anxious thoughts, and how to develop realistic thinking about anxiety-producing situations (Figure 4), (4) coping and relaxation skill activities with self-assessment of performance and rewards, (5) development of a hierarchy of feared situations and steps for gradual and repeated exposure to feared situations (using imagery and *in vivo* activities), (6) contingency management (examining the function of anxiety from a reinforcement perspective) and modeling (viewing videos of others confronting feared situations), and (7) skills for maintenance and relapse prevention. Animations, embedded video, audio playback, graphic novel style vignettes, image maps, timed prompts and on screen pop-ups provide an interactive and multimodal experience to promote sustained engagement with the treatment content across modules.

Adolescents are instructed at the beginning of the trial to use the program weekly. Adolescents who do not log in for one week receive an e-mail encouraging them to log in to complete their weekly module. During the program, adolescents have access to an *Ask the Expert* feature where they can e-mail a trained research team member with questions regarding the program and/or treatment. *Breathe* successfully underwent iterative usability testing cycles with five clinicians and five adolescents prior to the pilot study to ensure adherence to CBT therapeutic principles, developmental appropriateness for the target age group, satisfaction, and engagement with the multi-media content.

Figure 2. A screenshot of the Breathe program home screen.

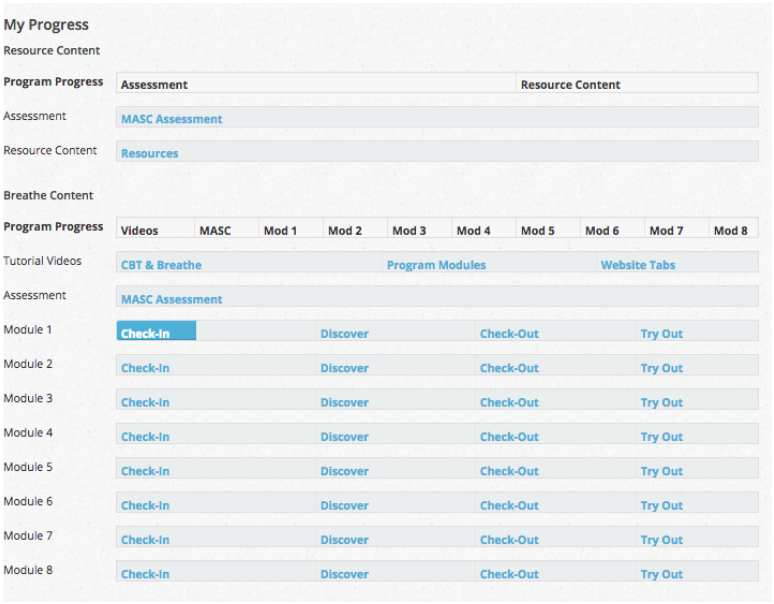


Figure 3. A screenshot of a weekly self-assessment activity.

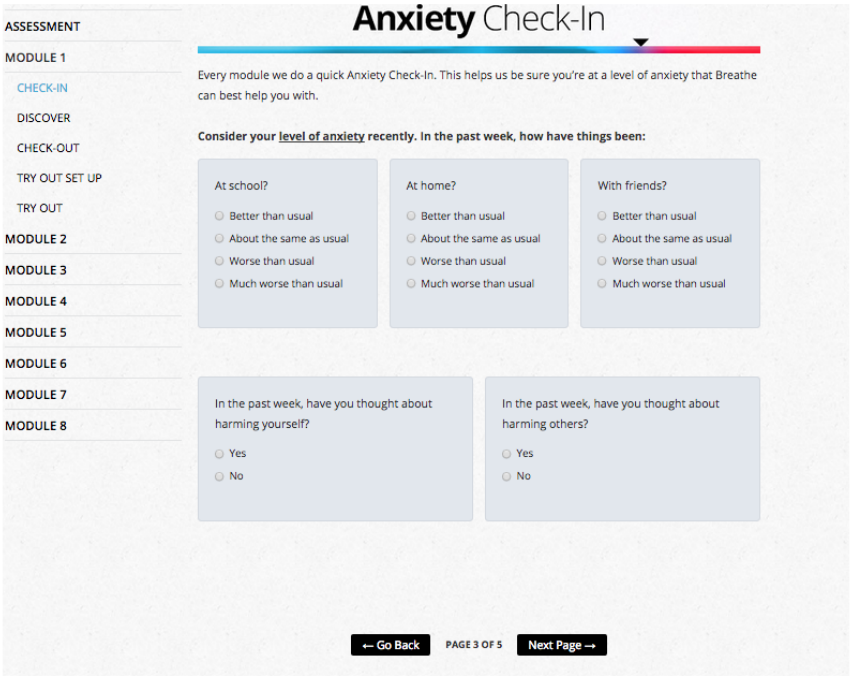
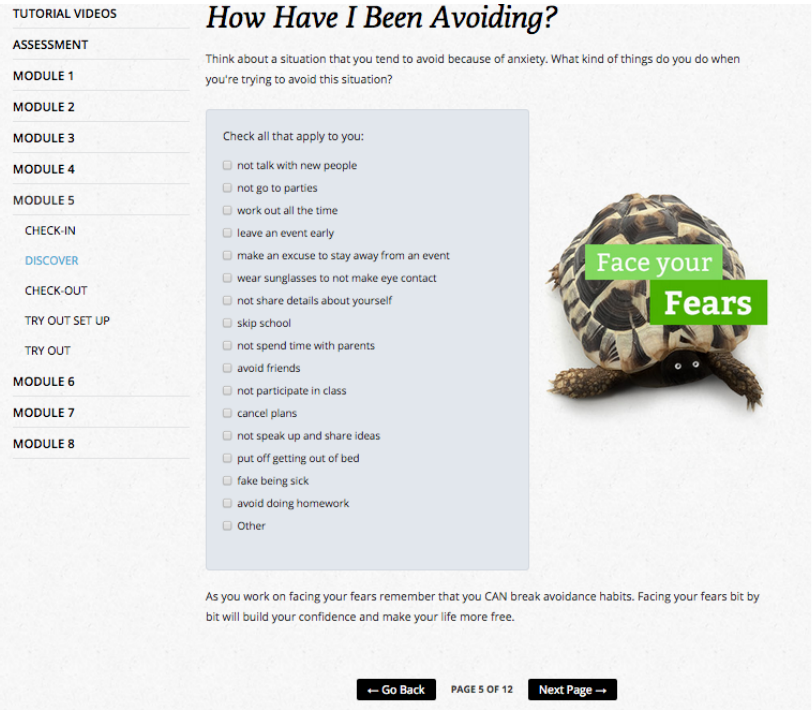


Figure 4. A screenshot of a learning activity.



Persuasive Design Mechanisms

Breathe is delivered via the Internet-based platform IRIS, which supports the integration of persuasive design mechanisms [50] for Internet-based interventions that are proven to encourage user engagement. Through the adaptive informatics architecture of IRIS, *Breathe* personalizes the program for adolescents via three primary mechanisms: *tailoring* content (ie, providing custom information and feedback to the adolescent based on their actions); *self-monitoring* (ie, enabling the adolescent to track their own behavior toward intended outcomes); and automated *suggestions/reminders* (ie, providing adolescents with information at the right time and the right context to help them keep on track). For example, an adolescent who indicates issues with alcohol or other drug use would receive material on these topics, while an adolescent who indicates alcohol or other drug use is not an issue would not. *Breathe* also engages the adolescent's parent(s) by sending e-mails with educational materials about the nature of adolescent anxiety and highlights of key topics their child is working on in the program. Adolescents are given the choice as to whether they want parents to receive these e-mail updates.

Safety Monitoring

The risks for adolescents participating in this pilot trial are considered minimal, but those in the treatment group are monitored each time they start and finish a module in the intervention program, to assess clinical functioning. Adolescents complete a *check-in* and *check-out* whereby they answer questions related to anxiety symptoms and indicate whether they are having thoughts of self-harm or harming others. This *check-in/check-out* is a typical clinical feature of CBT programs. The research team monitors the adolescents' answers through automated indicators built into the IRIS program. If IRIS flags a safety issue (eg, decompensation in anxiety symptoms, thoughts of self-harm), a research team member contacts the adolescent and their parent(s) by phone follow-up within 36 hours and directs them to emergency services if the adolescent requires more immediate care. Additionally, the adolescent is automatically directed onscreen to a safety video on self-harm and a pop-up box appears in the program encouraging the adolescent to notify a parent/guardian of their thoughts and to seek immediate help. Access to the *Ask the Expert* feature in *Breathe* and e-mail contact also allow adolescents to identify distressing issues that may be activated during treatment. Such adolescents are re-assessed to determine if continuing in the study is advisable. Those adolescents not able to use the program independently or who are struggling with additional mental health issues may be referred to a higher level of care. Any serious adverse event will be reported to the institutional ethics board according to the standard operating procedures. Adolescents randomized to the control group are provided with contact information for local emergency resources (crisis lines, emergency department and/or other crisis mental health resources).

Control (Minimal Intervention) Condition

Adolescents assigned to the control group are provided with access to a static webpage that houses suggested anxiety-related trade publications, print-based workbooks for adolescents, and the names of organizations and websites where the adolescent might find support. There is no interactivity or personalization included in the webpage. Adolescents assigned to the control group are provided with the option to use the *Breathe* program at the end of their 8-week control group participation. At that time, the study will follow the same processes to monitor safety and support adolescents who may require additional help, as detailed above.

Statistical Analyses

Adolescent reports of anxiety, intervention acceptability and adherence, use of co-interventions, recruitment and retention

rates, and demography will be described by numerical summaries (mean, frequency, range, and standard deviation [SD]). The mean difference in primary outcome change scores (and SDs) from baseline to the completion of module 8 (post-intervention) will be calculated with 95% CIs. SD values from the primary outcome change scores will be used to guide the full-scale RCT sample size calculations [51]. Adolescent global ratings of change (within the ranges of +2 to +3 or -2 to -3 for reported change using the 10-point Likert scale) will serve as the estimate for the MCID value [52]. The study recruitment rate (number of adolescents enrolled during the study recruitment period) and the retention rate (number of adolescents retained at the 3-month follow-up as indicated by MASC2 completion) will be calculated. These rates will be used to determine the number of study sites and time period needed for a full-scale RCT. Frequencies and proportions (with 95% CIs) will be used to describe categorical variables (eg, gender). We will summarize characteristics of completers (>75% of modules) and non-completers, and conduct tests of association to identify particular adolescents (eg, by gender) that may benefit from additional measures to maintain engagement, and will explore potential confounders (eg, age, sex, co-morbidity) that may be adjusted for in the full-scale RCT. Mean costs (and SDs) will be calculated (with 95% CIs) using published recommendations [46,47]. To account for the amortization of intervention use after upfront costs, we will distribute the investment costs over a 3- to 5-year investment time duration using a 5% societal interest rate and using 3% and 0% rates in the sensitivity analysis. Costs for reported co-interventions (health care services) will be estimated using Alberta costing and using 2015-dollar values.

Results

Currently, adolescents are being enrolled in the study. Enrolment is taking place between March 2014 and February 2016; data collection will conclude May 2016. We expect that analysis and results will be available by August 2016.

Discussion

As current systems-based models of health care look to enhance local community resource capacity, stepped care options and improved access to mental health services for adolescents are needed. Recent systematic reviews have shown that self-led, computer-based and computer-assisted CBT are viable treatment approaches for adolescents, but suggest that maturation of the evidence base is needed [30,31,32]. This pilot RCT is an essential step to designing a robust RCT to evaluate the effectiveness of Internet-based CBT for adolescents with moderate to mild anxiety problems. As a first step, satisfaction and treatment adherence results will indicate whether we need to make any critical intervention changes prior to a full-scale RCT.

We will use our pilot RCT results to develop a protocol for a full-scale trial. This trial will be designed to test the effectiveness of the *Breathe* program compared to a web-based, minimal control intervention providing psychoeducation (static webpage) in reducing anxiety symptoms in adolescents with moderate to mild anxiety problems. The MCID, sample calculation, and co-intervention use determined from the pilot RCT will inform full-scale trial methodology. Our sample calculation will ensure the full-scale trial is adequately powered, and will address sample size limitations noted in the literature to date [30]. The use of the MCID in the full-scale trial may also help to clarify inconclusive self-rated treatment effects reported across similar trials [30]. Our cost-consequence analysis will inform a cost-effectiveness analysis in the full-scale RCT. Finally, we will use the recruitment and retention rates calculated in the pilot trial to determine the number of study sites and timeline necessary to recruit the full-scale RCT sample.

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Authors' Contributions

ASN and PM conceived of the study, acquired funding, and participated in its design and coordination and helped to draft the manuscript. LW participated in the drafting of the study protocol and drafted the manuscript. AB, SC, EF, and MY participated in the design of the study related to clinical processes and safety monitoring, and will contribute to study monitoring. MJ and DWJ participated in the design of the study and multi-site trial management, and will contribute to trial monitoring. RJR and AO participated in the design of the study and will contribute to statistical and cost-consequence analyses, respectively. AJ participated in the design of the study related to measurement outcomes. All authors will participate in results interpretation and full-scale trial planning, and have read and approved the final manuscript.

Conflicts of Interest

The IRIS platform used in this study was developed by co-author Patrick McGrath and is currently being redesigned for commercialization.

Multimedia Appendix 1

Peer reviewer comments from the CIHR grant supporting this study. This manuscript reports Phase 3 (the pilot RCT) that is commented on in the reviews.

[PDF File (Adobe PDF File), 177KB - resprot_v5i1e18_app1.pdf]

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Abbreviations

ASQ: Ask Suicide-Screening Questions
Breathe: Being Real, Easing Anxiety: Tools Helping Electronically
CBT: cognitive behavioral therapy
IRIS: Intelligent Research Intervention Software
MASC: Multidimensional Anxiety Scale for Children
MCID: minimal clinically important difference
RCT: randomized controlled trial
SCARED: Screen for Child Anxiety Related Emotional Disorders
SD: standard deviation

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Protocol

Development, Validation, and Implementation of an Innovative Mobile App for Alcohol Dependence Management: Protocol for the SIDEAL Trial

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Abstract

Background: Information and communication technologies (ICT) have become one of the main pathways to the new paradigm of increased self-management of chronic conditions such as alcohol dependence. Validation of some mobile phone apps has begun, while validation of many others is forthcoming.

Objective: To describe the protocol for validation of a new app called SIDEAL (an acronym of the Spanish name "Soporte Innovador a la persona con DEpendencia del ALcohol," or innovative support for people with alcohol dependence).

Methods: The project consists of 3 complementary, consecutive studies, including a pilot feasibility study, a qualitative study using focus groups, and, finally, a randomized controlled trial where patients will be randomized to standard treatment or standard treatment plus SIDEAL. During the pilot study, feasibility, usability, and acceptance by users will be the main outcomes explored. An electronic questionnaire will be sent to patients asking for their opinions. Focus groups will be the next step, after which improvements and refinements will be implemented in the app. During the final phase, consumption variables (heavy drinking days per month, mean standard drinks per day) will be investigated, in order to test app efficacy.

Results: Because of the encouraging results with previous similar apps, we expect patients to widely accept and incorporate SIDEAL into their therapeutic options. Significant reductions in drinking-related variables are also expected. The pilot study has concluded with the inclusion of 29 patients. Results are expected to be available soon (expected mid-2016).

Conclusions: SIDEAL may represent a useful, reliable, effective, and efficient tool to complement therapeutic options available to both patients and professionals.

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KEYWORDS

alcohol dependence; alcoholism; telemedicine; mobile applications; eHealth; adherence; patient compliance; consumption register

Introduction

According to the World Health Organization [1], alcohol is the third largest contributor to the global burden of disease in developed countries, accounting for massive costs at both the individual and society level. Alcohol dependence, now formally labeled severe alcohol use disorder, is a medical, chronic, relapsing disease. In Spain, for example, it accounts for nearly 75% of the total burden of disease attributable to alcohol [2]. Because of its chronic, relapsing-remitting nature and high costs, there is a strong need to develop new ways to provide continuous care.

Information and communication technologies (ICT) could offer an approach to the addiction field through a new paradigm of increased self-management. Moreover, technology has the potential to provide continuous personalized care at a fraction of the cost of standard care. Existing evidence supports the suitability of chronic diseases as a target for technological assistance, to improve quality of life and reduce health care costs [3]. These facts have not gone unnoticed by national and international institutions, which are now clearly supporting and encouraging the development of such technologies. A clear example is the Horizon 2020 initiative of the European Commission, with an approximate budget for ICT-related projects of €560 million.

Intelligent mobile phones, or *smartphones* are currently widely used in Spain (4). Besides the usual telephone service, they offer computer-like functions with advanced software capabilities. Their portability, ease-of-use, and ubiquity make them excellent tools to enhance the management of chronic conditions. A new horizon of interactions between health care services and users with mobile phones as mediators can be envisioned. The range of possibilities includes scheduling visits for psychoeducation, monitoring, brief interventions, and biometric assessments.

A multitude of mobile apps specifically focusing on alcohol are currently available in the software market. However, recent reviews point out that a majority of them encourage drinking, while a smaller number support the reduction of problematic drinking. However, little research supporting their use is available [4,5]. One randomized, controlled trial was conducted, and encouraging results were reported [6]. Additional trials are expected in the near future, in a growing effort to apply scientific standards to this continuously and rapidly changing new area of ICT.

Recent publications [4,7-9] identified two key elements critical to the efficacy of apps in the management of chronic diseases. These include ongoing monitoring and use of push technology (which provides sustained proactivity). Other success-conditioning factors are usability, institutional support, expert endorsement, and individualized adjustment or tailoring. It is also strongly urged that these apps be evaluated using the scientific method, which will allow for the implementation of apps based not only on their usability, but also on their validity, effectiveness, and efficiency.

Mobile phones have been suggested to be suitable platforms for the management of alcohol use disorders via apps that integrate different functions. These include skills training, motivation enhancement, psychoeducation, social support, relapse prevention, and reminders (medical visits, medication schedules, health events). In addition, technology allows for the implementation of a system for continuous monitoring and ecological momentary assessment ("in the moment") [4]. The information gathered can then be displayed to the user (thus providing feedback) in an attractive way, facilitating modulation of the problematic behavior. All these features and functions therefore allow not only for simple and efficient collection of information, but also for the combination of this information with other functions to potentially encourage motivation, decrease problems associated with consumption, and even support relapse prevention [10].

The SIDEAL project (an acronym of the Spanish name "*Soporte Innovador a la persona con DEpendencia del ALcohol*," or innovative support for people with alcohol dependence) was undertaken in this context. SIDEAL is a public-private collaboration of Lundbeck Spain SA, Pulso Ediciones, SL, the Subdirectorate of Drug Addictions of the Catalonia regional government, and the Addictions Unit of the Clinic Hospital of Barcelona. A Web-based app system has been developed to support alcohol-dependent patients. This paper reports its theoretical basis and development process, as well as the protocols of the different studies that will be conducted to validate its usability and efficacy.

Methods

Theoretical Basis and Patient Implementation

The main source of theoretical guidance for SIDEAL comes from motivational interviewing (MI) principles [11]. MI is intended to help people move through the stages of change [12] in a nonjudgmental, facilitative manner. Although it has a directive, goal-oriented style, it is respectful of patients' goals. MI also encourages an individual, tailored approach for each patient. Therefore, SIDEAL is respectful of patients' goals, and allows for tailoring of the app according to their preferences. Other principles adopted in the development of the app include design simplicity, noninvasive proactivity, privacy, professional endorsement and support, and adjustment to individual needs. An increasing number of guidelines and recommendations for developing apps aimed at behavioral change [13,14] are also currently available and continue to be useful as a source of practical guidance.

While other apps currently being validated, such as the Location-Based Monitoring and Intervention for Alcohol Use Disorders (LBMI-A) [8], have been designed as stand-alone, self-administered, independent treatments for alcohol patients, the purpose of SIDEAL is to become a tool that both patients and professionals can incorporate into their therapeutic armamentarium. Although the app may work as a stand-alone option, we think that it is best used under the principles of shared information and decision-making between patients and professionals. All information produced by users is transferred to a Web system to which professionals have access. In order to respect patients' privacy, however, users always have to proactively allow the information to be transferred to the Web by clicking a specific button in the app.

The app is currently available free of charge in the main app markets (ie, Apple Store and Google Play). However, to create a user account and tailor and personalize features, patients should have an initial visit with a professional, during which both the patient and professional will configure the user's account via the Web system. For example, the main treatment goal will be set (choosing between complete abstinence and reduction). If reduction is selected, a desired daily consumption limit will also be set. If a patient is taking any specific medication, this will also be recorded in the user's account.

Based on patient preferences, professionals will be able to select which psychoeducational text message (short message service, SMS) the user will receive on a weekly basis.

SIDEAL Development Process

The development and testing phases of the Web-based app system were conducted from September 2013 to December 2014 by a collaborative team of specialists in addictions (psychiatrists) and software engineers, as well as graphic designers and project managers, with previous experience in the development of patient support software. All user requirements, functions, and software design were first established, and the installation (soft and hard), operational key functions, and performance requirements were subsequently tested to evaluate the entire system.

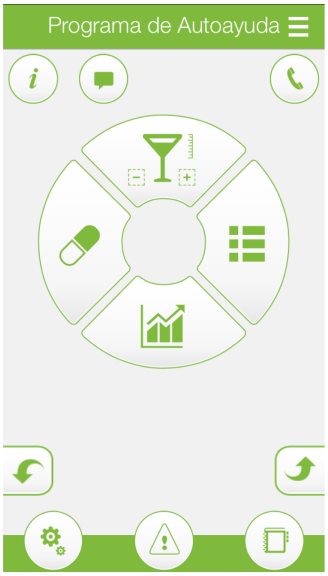
In addition to compliance with security and privacy rules, relevant app requirements and functionalities include easy-to-use functions for selecting a program (reduction or abstinence); recording daily alcohol consumption (standard units); graphically monitoring standard units in reference to the established limit; accessing educational information; and displaying a telephone help line (public or private). For users monitored by a physician who previously created a user profile for them through the Web interface, additional app functions were included for downloading consumption goals and prescribed therapies from the dedicated professional Web interface; recording and monitoring treatment adherence; receiving automated personalized feedback (weekly via the app); receiving one general piece of advice weekly (a text message selected at random from a pool of 50); and receiving surveys for users to provide feedback.

All 50 text messages were selected and reviewed by the investigator's panel based on best practice standards and consensus. Updated published educational information was used.

SIDEAL Functionalities

The different "modules" or functionalities included in the app may be accessed in parallel, with no restrictions, in contrast to the stepwise approach implemented in other apps. Figure 1 shows the initial screen, where all functionalities may be accessed.

Figure 1. Initial screen.



Consumption Records

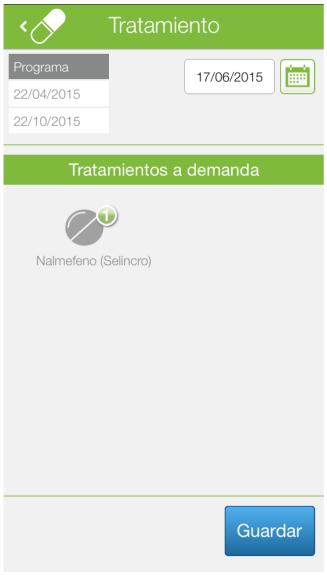
Patients record alcohol consumption using icons representing alcoholic drinks. The system automatically converts this information into standard units and displays it in a graphic where the agreed limit is also displayed. The user can set reminders that will appear when consumption limits are exceeded. Consumption may be introduced gradually throughout the day, all at once, or even on subsequent days using the calendar function, which allows for the selection of previous dates. The system also allows patients to record that no consumption has occurred during the day. Figure 2 shows a consumption register screen.

Figure 2. Consumption register.

Drug Adherence Record

If the patient is receiving an alcohol-specific drug treatment, the system also allows for recording daily drug compliance. The system automatically computes percent drug adherence. Figure 3 shows one of the screens of this module.

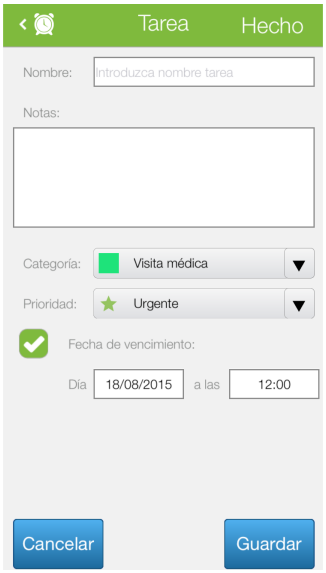
Figure 3. Pharmacological adherence register.



Calendar

Patients can enter the dates of visits to different health care professionals. The system will produce reminders with push technology to remind patients of their scheduled visits. Figure 4 presents a screen showing this function.

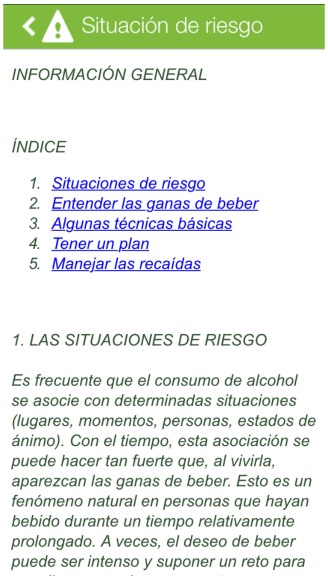
Figure 4. Calendar.



Medical Information and Psychoeducation About the Disease

Basic information about alcohol use disorders is presented, as well as links to useful, trustworthy websites. A special and separate body of information on risky situations and craving is included (see Figure 5). In addition, an SMS text message with psychoeducational content is sent weekly to the user.

Figure 5. Psychoeducation module.



Weekly Questionnaires

Weekly surveys are sent to patients via push technology, including a questionnaire on satisfaction with their general progress, and another regarding their craving for alcohol. Both employ Likert scales and use the previous week as a reference.

Weekly Feedback

Once a week, via push technology, patients receive a short feedback message of a motivational nature about their use (number of days during the last week) of the consumption recording module.

Validation Process and Methodology

We will carry out 3 successive studies to evaluate and validate SIDEAL. All studies will recruit adults (≥18 years of age) from the outpatient clinic of the Addictions Unit of the Clinic Hospital of Barcelona. Recruitment will follow a consecutive sampling strategy. Subjects will be required to sign an informed consent form before they are recruited into any study. They will also be required to have a mobile phone with an active Internet connection. Exclusion criteria for all studies will be addiction to any other substance (excluding nicotine), cognitive decline, technological illiteracy, or any other condition precluding the use of a mobile phone.

Feasibility Study

A 6-week pilot study will first be conducted to assess SIDEAL feasibility and usability, and users' satisfaction. As we will have equipped the app with all features suggested in the literature to maximize feasibility, usability, and satisfaction, we expect high scores on outcome variables in this first stage. In this regard, personalization, user engagement (via push technology and weekly feedback reports), reduced invasiveness and enhanced privacy, and a high-quality user interface were of critical importance in the development of the app, and are the expected drivers of the feasibility study outcomes.

Because of the pilot nature of the study, 30 patients are considered an adequate sample size. Therefore, 30 outpatients with alcohol use disorders will be given the opportunity to install the app on their mobile phone, and will be asked to use it for 6 consecutive weeks. Inclusion criteria will be adults suffering from alcohol dependence according to DSM-IV or from severe alcohol use disorder according to DSM-V (as clinically diagnosed by their psychiatrists).

At baseline, sociodemographic data (age, sex, marital status, education level) will be collected. Clinical variables related to the disease and drinking status will also be collected at baseline and at study end, and will be taken from the Timeline Follow-Back (TLFB), which will be completed for the preceding 6 weeks at both baseline and study completion. The main variables analyzed will be the number of heavy drinking days and mean alcohol consumption in units per day. When the 6-week period is over, patients will be asked to complete an electronic questionnaire on app usability and satisfaction. The questionnaire will be an adaptation of the Usefulness, Satisfaction, and Ease-of-use Questionnaire (USE Questionnaire) [15] and will consist of 30 items to be rated by users on a 7-point Likert scale (from 0 to 6). Items are grouped into 4 main categories: usefulness, ease of use, ease of learning, and satisfaction. Extra questions about which modules were found to be most useful, and a free-text section to allow patient comments on any point they consider relevant, will be added to these items. The questionnaire data will be reported according to the Checklist for Reporting Results of Internet E-Surveys (CHERRIES) checklist [16]. Complementary data regarding usability, alcohol consumption, craving modulation, and other clinical variables will also be collected from the data generated by the app.

Statistical analysis will consist of a descriptive analysis of sociodemographic and clinical variables. Acceptance and feasibility will be analyzed as percentage of days used and through the satisfaction questionnaire, for which results will be analyzed using means and standard deviations of all respondents for each category of the questionnaire. The proportions of patients reporting high, medium, or low levels of satisfaction will also be calculated. Paired *t* tests and chi-square tests will be used for analysis of drinking variables (difference from baseline to study end). These variables will be the number of heavy drinking days and the mean number of units per day. Because of the exploratory nature of the pilot study, no regression analysis will be conducted.

Focus Groups

Two different single-session focus groups (one with patients, one with psychiatrists) will be carried out to discuss the experience with SIDEAL, and to collect suggestions and reports of errors. A maximum of 10 subjects included in the feasibility study will be selected for the focus group.

Personal interviews with some users will also be conducted to collect qualitative information. Data will be analyzed using qualitative methods. Focus group sessions will be transcribed verbatim and analyzed with NVivo 10 or a similar software package. In analyzing the transcripts, areas of consensus and discrepancy will be discussed. Coding and categorizing will follow the principles of inductive content analysis [17]. The information collected will be helpful to improve SIDEAL, a new version of which will be produced if considered necessary.

Randomized Controlled Trial

The last step will be the completion of a randomized, controlled trial. Selected subjects will be randomized to standard treatment plus SIDEAL or to standard treatment alone. There will be no exclusion criteria with regard to what is considered standard treatment. In this sense, patients may be receiving any treatment, pharmacological or psychosocial, on an individual or group basis. The study will last 24 weeks. The initial hypothesis, based on previous research on the efficacy of alcohol apps, is that the addition of SIDEAL will improve drinking outcomes in alcohol-dependent patients, with especially significant reductions in the number of heavy drinking days and mean alcohol consumption. User acceptance and usability will be essential to achieve this objective. In addition, continuous monitoring and feedback, as well as psychoeducational content and messages, are expected to be effective for decreasing alcohol consumption.

To be included, subjects will be required to have a diagnosis of alcohol dependence or alcohol use disorder according to their psychiatrists, a drinking pattern in the previous 4 weeks consisting of >5 heavy drinking days (HDD; defined as a day with alcohol consumption ≥60 g for men and ≥40 g for women), and an average alcohol consumption above medium risk levels (ie, >40 g of alcohol/day for men and >20 g of alcohol/day for women) in the 4 weeks before study entry. Patients will be evaluated at baseline and week 24. Baseline assessments will be the same as those in the feasibility study, with the addition of the following liver enzyme tests: gamma-glutamyl transferase (GGT), aspartate aminotransferase (AST), and alanine aminotransferase (ALT); and the Addiction Severity Index (ASI) [18]. At week 24, drinking variables (number of HDDs and mean units per day) taken from the TLFB for the previous 6 weeks will be collected. Blood tests and the Addiction Severity Index will be evaluated again.

The statistical analysis will consist of a multivariate regression analysis with change from baseline to study end in number of HDDs and mean alcohol consumption per day as the dependent variables. Age, sex, baseline alcohol drinking, and ASI scores will be entered as independent variables. A secondary analysis will compare changes between groups in ASI scores and liver enzyme levels. A logistic regression analysis will also be

conducted with the same independent variables, where rates of point and continuous prevalence of abstinence will be considered as the dependent variables.

For sample size calculation, using a significance level of 5%, and assuming a standard deviation for the change from baseline in number of HDD of 6 days, 45 patients in each treatment group would provide 80% power for detecting a difference between treatment groups of 4 heavy drinking days, assuming a dropout rate of 25% at week 24. Results will be reported according to the CONSORT-EHEALTH checklist [19].

Results

The pilot study has concluded with the inclusion of 29 patients. Results are expected to be available soon.

Discussion

Improved care, decreased cost, increased efficiency, and strengthened communication between patients and professionals are the advantages promised by mobile technology in the addiction field. A new paradigm of self-management for chronic conditions such as addiction is growing closer.

We hope that validation of this app will help patients improve the management of their condition, increasing treatment efficacy and reducing costs. There is clear support for this process from both public and private institutions, a fact that we consider essential to ensure wide and firm implementation.

We expect the app to be flexible. This means that the app will surely be improved during the validation process, and other functionalities or modules may be incorporated. Patient feedback will be indispensable for this task.

Recent studies validating similar apps for patients with alcohol disorders have reported promising results. For example, Gustafson et al [6] recently reported the results of a randomized, controlled trial of the Addiction-Comprehensive Health Enhancement Support System (A-CHESS) app, for which a significant reduction of risky drinking days was found in patients receiving the app. Dulin et al [15] reported the results of a pilot study with the app LBMI-A. Although this was an exploratory study, results were also encouraging, with patients reporting high degrees of usefulness and also showing significant reductions in hazardous alcohol use while using the system.

The complex pathophysiology of alcohol use disorders, where both psychosocial and biological factors are intertwined, should not be forgotten. In alcohol-dependent patients, many resources and treatment components are usually needed to achieve stable remission or an enduring reduction. Hence, we think that expectations of mobile technologies applied to the field of alcohol addiction should be accordingly reasonable. In this regard, we expect SIDEAL to become an add-on to the different options available to professionals and patients when devising a therapeutic plan.

A main limitation of this project is the fact that many alcohol-dependent patients may have some degree of cognitive decline [17,18], which could prevent them from using the app. Although, as previously stated, we expect the app to become a common tool for patients and therapists, there remains a low chance that patients will decrease their attendance at face-to-face visits. Other methodological concerns are the impracticality of blinding participants to study interventions and the fact that patients will be recruited from the same site, which may decrease the external validity of the study findings.

As a conclusion, we expect to add scientific knowledge and apply rigor to the ever-evolving field of mHealth. In this process, we expect SIDEAL to become a reliable, useful, and effective tool for alcohol-dependent patients and their health care professionals.

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Authors' Contributions

AG and PB were responsible for the study design and for writing the manuscript. AG was responsible for coordination with the relevant companies participating in the project. XB participated in design and testing of the Web-based app system and in writing the corresponding parts of the manuscript. LO participated in both study design and writing of the manuscript, making substantial contributions to its final version. All authors read and approved the final manuscript.

Conflicts of Interest

Dr Gual received research grants from Lundbeck SA, D & A Pharma, and TEVA, and honoraria from Lundbeck SA, D & A Pharma, and Abbivie. Dr Pablo Barrio received honoraria from Lundbeck SA. Dr Ortega has no competing financial or personal interests. Xavier Bona is an employee of Pulso Ediciones, the company developing SIDEAL.

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Abbreviations

ALT: alanine aminotransferase
ASI: Addiction Severity Index
AST: aspartate aminotransferase
A-CHESS: Addiction–Comprehensive Health Enhancement Support System
CHERRIES: Checklist for Reporting Results of Internet E-Surveys
GGT: gamma-glutamyl transferase
ICT: information and communication technologies
TLFB: Time-line Follow Back
USE Questionnaire: Usefulness, Satisfaction, and Ease of use Questionnaire

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Protocol

Exploring a New Simulation Approach to Improve Clinical Reasoning Teaching and Assessment: Randomized Trial Protocol

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Abstract

Background: Helping trainees develop appropriate clinical reasoning abilities is a challenging goal in an environment where clinical situations are marked by high levels of complexity and unpredictability. The benefit of simulation-based education to assess clinical reasoning skills has rarely been reported. More specifically, it is unclear if clinical reasoning is better acquired if the instructor's input occurs entirely after or is integrated during the scenario. Based on educational principles of the dual-process theory of clinical reasoning, a new simulation approach called simulation with iterative discussions (SID) is introduced. The instructor interrupts the flow of the scenario at three key moments of the reasoning process (data gathering, integration, and confirmation). After each stop, the scenario is continued where it was interrupted. Finally, a brief general debriefing ends the session. System-1 process of clinical reasoning is assessed by verbalization during management of the case, and System-2 during the iterative discussions without providing feedback.

Objective: The aim of this study is to evaluate the effectiveness of Simulation with Iterative Discussions versus the classical approach of simulation in developing reasoning skills of General Pediatrics and Neonatal-Perinatal Medicine residents.

Methods: This will be a prospective exploratory, randomized study conducted at Sainte-Justine hospital in Montreal, Qc, between January and March 2016. All post-graduate year (PGY) 1 to 6 residents will be invited to complete one SID or classical simulation 30 minutes audio video-recorded complex high-fidelity simulations covering a similar neonatology topic. Pre- and post-simulation questionnaires will be completed and a semistructured interview will be conducted after each simulation. Data analyses will use SPSS and NVivo softwares.

Results: This study is in its preliminary stages and the results are expected to be made available by April, 2016.

Conclusions: This will be the first study to explore a new simulation approach designed to enhance clinical reasoning. By assessing more closely reasoning processes throughout a simulation session, we believe that Simulation with Iterative Discussions will be an interesting and more effective approach for students. The findings of the study will benefit medical educators, education programs, and medical students.

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KEYWORDS

clinical reasoning; simulation; debriefing; iterative discussions; diagnostic errors; cognitive bias; verbalization; dual-process theory

Introduction

Background

The Importance of Clinical Reasoning in Medicine

Diagnostic errors account for more than 8% of adverse events in medicine and up to 30% of malpractice claims [1]. These errors may be related to the working environment but clinical reasoning issues are involved in about 75% of the cases, either alone or in association with system failures [2].

In this context, clinical reasoning is a crucial skill for all physicians regardless of their area of expertise and becomes a central aim of medical education [3]. The clinical reasoning process is largely supported by several decades of research in cognitive psychology and has been extensively published [4-6]. The most accepted model of clinical reasoning is called *dual-process framework* and consists of two independent systems [7]. System-1 is automatic, intuitive, nonanalytical, and error-prone. It leads to the immediate recognition of the clinical constellation and the generation of a working diagnostic hypothesis. System-2 is slower, more analytical, and conscious [8,9]. Novices employ this analytic mode of reasoning more frequently than their experienced counterparts because they lack the necessary experience for System-1 reasoning. Dual-process theory posits that both systems are simultaneously required in most clinical scenarios and has been associated with better diagnostic outcomes [10]. However, in what situations does the valence go towards one system or another remains unclear and how both systems are activated and used is still under study and debated [11-13]. Preliminary conclusions from recent publications describe that the analytical system is primarily used in the following situations [14,15]: when time permits, when there are high-stakes outcomes, when the situation is complex, when the decision-maker is facing ambiguous, nonroutine or ill-defined problems, and in the context of uncertainty. In contrast, routine problems associated with a higher level of certainty would be more often dealt with by the intuitive system, especially when time is lacking [11-13].

Another challenge for medical educators is the assessment of residents' clinical reasoning [3]. Because of the paucity of scientific evidence about optimal evaluation, both quantitative and qualitative clinical reasoning assessment tools have been reported [5,16-19]. However, the following general issues arise from these tools: (1) diagnostic reasoning must be inferred from behavior because it is not a discrete and measurable quality; (2) most of these instruments are performed in the classroom that emphasize the assessment of System-2, but not System-1 reasoning nor the shift between automatic and analytic reasoning [15,16]; and (3) rater's personal knowledge, experience, ability, and cognitive biases influence his or her adjudication of a learner's performance in a nonstandard fashion [20-24]. Moreover, reasoning assessment can be highly complex and dependent upon the context. Durning et al [25] have recently reported the influence of three environmental factors on clinical reasoning: (1) the patient's specific problem (patient factors); (2) the setting in which the patient is evaluated (encounter factors) [10,26]; and (3) human-factors such as fatigue, well-being, and sleepiness (doctor factors). They emphasize the importance of measuring the environment as a part of the signal, rather than part of the "noise", which is to be minimized and generally ignored [27-29].

Simulation-Based Education as a Strategy to Enhance Clinical Reasoning Skill

Simulation-based education (SBE) has recently emerged as an instrument with potential to assess diagnostic reasoning [16,30,31]. Based on Kolb's learning cycle [32], true learning is depicted as a four-part process in a cycle. Individuals learn through concrete experience (phase 1), reflection on the experience (phase 2), conceptualization of their reflective observations into more abstract models (phase 3), and experimentation of these new principles and conclusions to guide subsequent decisions and actions that lead to new concrete experiences (phase 4). Phase 2 and phase 3 are components of debriefing, a learning activity that generally follows a simulation experience. According to many authors, debriefing could provide an opportunity for residents and faculty to re-examine what occurred during the simulation process and detect possible flows in the reasoning process [33-45].

An important question remains whether exploration of the diagnostic process, by providing an opportunity for learners to reflect upon past clinical decisions, is more effective after the scenario or if it is integrated during the simulation session [45,46]. In the classical approach of simulation, debriefing follows the simulation experience. However, the educational valence of this type of session presents several limitations concerning the clinical reasoning assessment. First, this way of *reflection-on-action* means that it is mainly the analytical part of the reasoning process that is explored during the debriefing, without focusing on the intuitive process [47]. Second, after a stressful scenario, residents frequently forget or modify what they said or thought according to the evolution of the case, even if video recordings provide insight into what may not be documented in the medical record or fully observed in real time [45].

We believe that changes in the organization of a simulation session could allow better assessment of both System-1 and 2 of the reasoning process. First, *reflection-in-action* should be encouraged by concurrent verbalization to let the tutors know about the student's intuitive System-1 thinking during management of a patient [11-13]. Second, *reflection-on-action*, which reflects analytical System-2 should be encouraged by in-simulation interruptions at key moments of the clinical reasoning process [48,49]. These interruptions could be assimilated to a dynamic and decision-dense environment where clinical reasoning constructs must be considered, as studies suggest that the average time on particular tasks is limited to less than 2 minutes, and interruptions occur every 2 to 10 minutes in the emergency department [50-52]. Finally, the active experimentation of newly acquired conceptualizations in a subsequent part of the scenario may avoid the learner's return to actions based on habits and nonreflective experience [32]. Based on these arguments, we present a new approach of SBE, called simulation with iterative discussions (SID). The simulation session is designed as a single scenario with a computerized mannequin where the instructor interrupts the

flow of the scenario at three key moments to cue residents towards the appropriate medical management of the case. The objectives of the session are to build a scaffold of clinical reasoning competencies throughout the scenario while continuing to manage the patient, and to improve concept acquisition and retention with spaced learning. We believe that a closer assessment of both System-1 (by concurrent verbalization during patient management) and System-2 (during iterative discussions) will improve students' capability to self-improve clinical reasoning skills in an authentic setting.

Why Is It Applied in Neonatology?

Implementing a new educational strategy that assesses clinical reasoning should be particularly exciting in a busy clinical environment such as neonatology. In contrast to the well-established management of neonatal emergencies at birth [53], most daily clinical situations managed in neonatology are nonroutine or ill-defined problems marked by high levels of complexity and unpredictability, requiring that clinical reasoning is solely based on the analytical System-2 [3,6,14,15,54]. Moreover, the frequency and the impact of diagnostic errors and cognitive biases increase in emergency settings due to several factors such as stress, fatigue, circadian disruptions, time constraint, and noisy environment. In these high-risk situations, physicians rely on System-1 process [55-58]. By enabling both System-1 and 2 of clinical reasoning, practicing neonatology requires robust clinical reasoning abilities in addition to cognitive, technical, and behavioral skills.

Aim of the Study and Working Hypotheses

The aim of this study is to explore how clinical reasoning abilities of residents in General Pediatrics and Neonatal-Perinatal Medicine evolve and are learned with the SID in comparison to the classical approach of simulation.

We hypothesize that (1) SID allows better assessment of both System-1 and 2 of clinical reasoning; (2) SID promotes higher self-progression of the clinical reasoning process when compared to the classical approach of simulation; (3) concurrent verbalization benefits mainly novice residents with underdeveloped System-1 reasoning process; and (4) iterative discussions benefit both novice and expert residents by enhancing System-2 processes.

Methods

Setting and Population

The study will take place at the Mother-Child Simulation Center at CHU Sainte-Justine, between January and March 2016. CHU Sainte-Justine is a standalone pediatric center that houses a 65 bed level 3 Neonatal Intensive Care Unit (NICU). The simulated setting will be a NICU. The simulation center is equipped with appropriate audio-visual equipment.

Residents enrolled in the General Pediatrics and Neonatal-Perinatal Medicine programs will be the target population for this study. This population is selected because (1) residents are regularly exposed to simulation training during their curriculum, and (2) their clinical reasoning ability improves between the 1st and the 6th year of residency, mainly with increasing clinical exposure through rotations [59]. All postgraduate year (PGY) 1 to 6 residents enrolled in the General Paediatrics and Neonatal-Perinatal Medicine programs at Université de Montréal between May 1 and June 30, 2015 will be eligible for inclusion. There will be no exclusion criteria.

Ethical Considerations

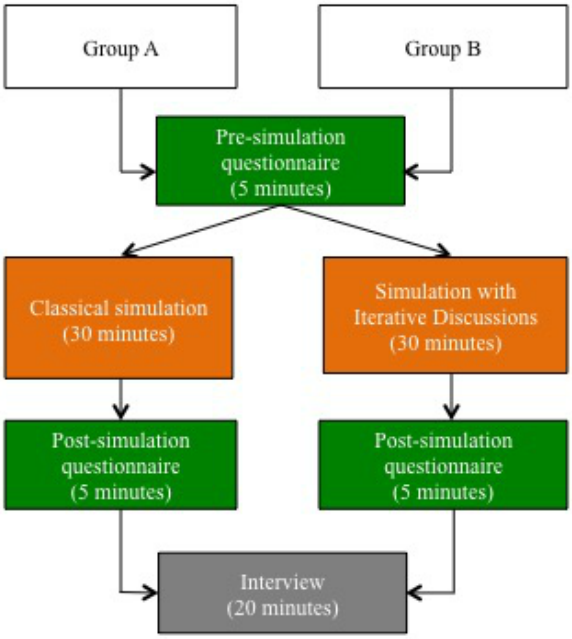
Each resident will be approached for consent by one of the authors. He/she will be informed that participation or lack of participation in the study will not impact residency training assessment. There will be no financial incentive to participate, and participants will be able to opt out at any moment of the study. The findings of this study will be treated anonymously. The project has been approved by CHU Sainte-Justine's institutional review board on July 15, 2014.

Study Design

This is a prospective exploratory nonblinded randomized mixed-methods study. Both quantitative and qualitative research methods will be used simultaneously as to comprehensively explore how clinical reasoning develops through both simulation modalities.

Randomization will be stratified according to two different groups depending on their level of exposure to the NICU: (1) novice residents (PGY1 and 2, exposed to less than 8 weeks to the NICU), and (2) expert residents (PGY3-6, exposed to at least 8 weeks to the NICU). These residents have been respectively exposed to at least 5 or 15 simulation sessions during their residency training. Residents will be randomly (by draw of names) allocated by the primary investigator to group A or group B (Figure 1). Group A will be exposed to the SID approach, whereas group B will be exposed to the classical approach of simulation. Participants will be scheduled to complete the study protocol in 60 minutes. In the first 5 minutes, participants will complete the presimulation questionnaire and receive a brief introduction including a period to get physically familiarized with the mannequin (SimNewB; Stavanger, Norway). In the next 30 minutes, residents will complete the clinical simulation scenario after reading a clinical vignette. At the end of the session, participants will have 5 minutes to complete the postsimulation questionnaire (see Multimedia Appendix 1). The course will end with a 20 minutes semistructured interview.

Figure 1. Study protocol.



Intervention

Personnel

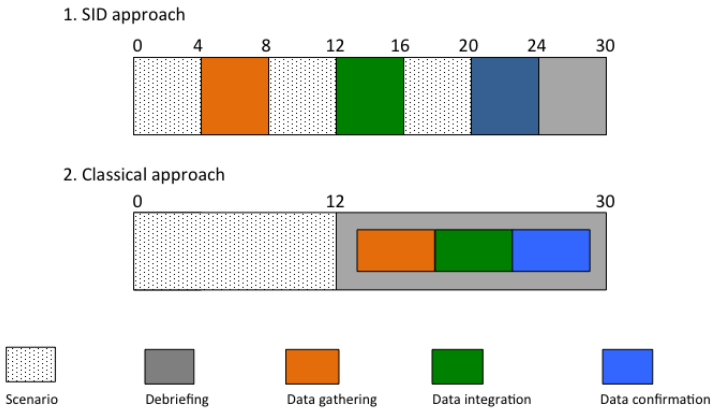
An instructor experienced with programming, control of the computerized mannequin, and the art of debriefing will be in charge of running the scenario according to the residents' actions and will conduct the iterative discussions and debriefings. A facilitator will also be present in the simulation room to ensure the flow of the scenario and will provide necessary information about the case upon request from the resident. Standardized health professionals (respiratory therapist and/or a nurse working in the Sainte-Justine hospital NICU) will be present and will portray a specialist from their proper field.

Description of the Intervention

The SID approach (Figure 2a) consists of a scenario interrupted at three moments and followed by a short debriefing. According to Kuhn's steps of the medical reasoning process [60], the session is divided into "data gathering", "data integration," and "data confirmation". Each part consists of two phases. First, the participant is asked to manage a simulated patient based on a real-life and complex case. Complexity, uncertainty and environmental factors such as doctor, patients, and encounter factors [25] are voluntarily embedded. The participant is also asked to verbalize his or her first *intuitive* diagnosis as it comes in mind by System-1 activation. Second, the scenario stops and the instructor questions the participant on his clinical reasoning process at that point in time by System-2 activation. Each interruption must be as short as possible in order to keep the trainee in action, and is ended by a one-sentence reconceptualization of the scenario by the instructor before pursuing the scenario (for the two first stops). Discussions include questions regarding data gathering and the rationale of ordered investigations (first stop), data integration, and how investigation results helped reach a diagnosis (second stop), and finally data confirmation and how the management decisions were reached (third stop). There is neither feedback nor guidance from the instructor during the stops in order to not interfere with the participant's ongoing clinical reasoning process. These stops are "discussions" and not "debriefings". A short general debriefing ends the session, and provides feedback on reasoning, procedural skills, and knowledge by highlighting the learnt key messages.

The classical approach (Figure 2b) consists of a scenario with no intervention provided by the instructor until debriefing. As in the SID approach, the participant must verbalize his reasoning process and the diagnostic hypotheses as they come to mind during the scenario. Immediately after the scenario, a facilitated debriefing is performed. This debriefing lasts two to three times the length of the scenario and focuses on the participant's clinical reasoning skills.

Figure 2. Simulation formats. This figure represents structures of (a) SID and (b) classical approach of simulation, with approximate timing in minutes. (a) A three-time interrupted scenario, with three stops that represent iterative discussions concerning data gathering, data integration, and data confirmation. There should be no guidance by the instructor until the real and short debriefing ending the session. (b) A one-shot scenario followed by a true debriefing conducted by the instructor according to the item checked in the clinical reasoning assessment tool. As a standard debriefing, retroaction from the instructor will be possible.



Clinical Reasoning Assessment Tool

The Clinical Reasoning Assessment Tool (Figure 3) aids the instructor to identify proper questions during the iterative discussions (SID approach) and in conducting debriefing (classical approach). The authors of the present study have developed this tool because of the absence of such a tool in the literature. It is based on Graber's classification of diagnostic errors [2,25], Audétat's practical guide [61] to assist clinical teachers in detecting clinical reasoning difficulties and follows Kuhn's steps of the medical reasoning process [60]. The objective of this tool is to help the instructor detect the student's type of diagnostic errors focusing on his reasoning process, and then to determine appropriate questions to ask the student in order to let him verbalize and possibly self-correct his reasoning process. Three types of environmental factors leading to diagnostic errors may be involved in the reasoning process: nonfault factors (also named patients errors, which are out of the control of the physician), human factors and cognitive factors (also named doctor errors) and system factors (also named encounters errors, due to organizational or institutional flaw). Specific failure in the doctor's cognitive process may be due to faulty knowledge, faulty recognition of cognitive biases or faulty data gathering (orange squares), data integration (green squares), and data confirmation (blue squares). One or two questions per section are suggested to the instructor to allow for exploration of each type of diagnostic error. Finally, a list of the main cognitive biases according to Croskerry et al [62] is provided and a suggested approach is presented. Once the type of error is identified, the instructor has to find one or two specific questions in order to let the student verbalize (or self-correct) his reasoning mistake.

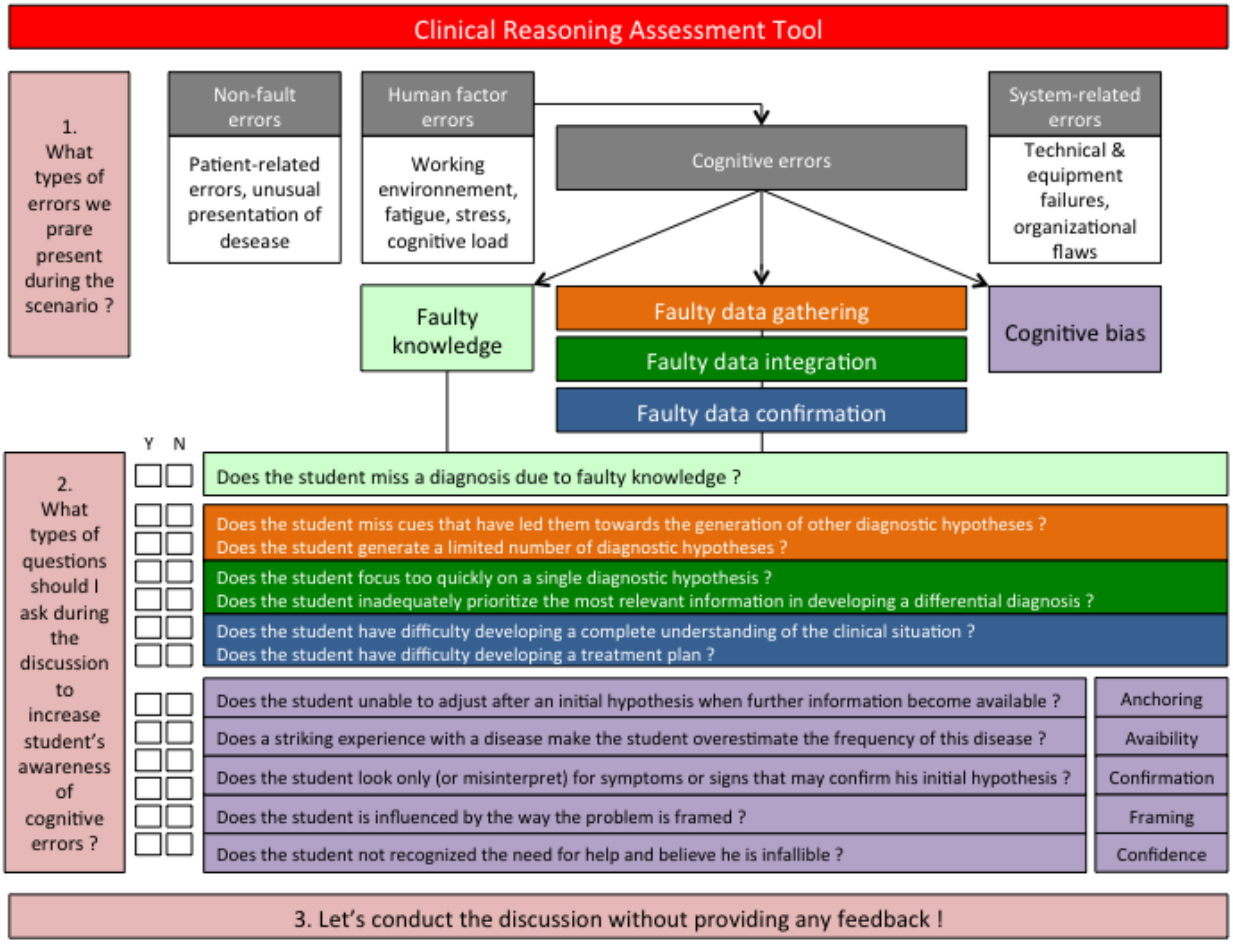
Scenario

In order to stimulate the reasoning process, the chosen topic has to be realistic, and hold a range of differential diagnoses. Investigations should be necessary to precise or refute diagnoses in the absence of specific clinical signs. Uncertainty needs to be deliberately embedded. Management must be complex with controversies concerning treatments. Moreover, a range of environmental factors and events can be added to challenge team members by generating dissonance and failures in order to optimize efficiency of simulated team training and adult learning. The amount of provided information has to be minimal and nonspecific aiming to stimulate additional questions from the participant. Answers to participants' questions must be standardized and should cover a range of possible differential diagnoses. Finally, procedural and relational skills must be embedded in order to portray as closely as possible the real-life environment.

The general structure of the scenario must follow three parts so it can be performed in a single run-through (for the classical approach) or interrupted (for the SID approach). First, a nonmonitored patient presents with minor symptoms but remains clinically stable. The participant has to check the vital signs, ask the nurse for the history and the results of the physical examination, and order investigations. Second, the patient presents with acute collapse. The participant has to interpret investigation results while managing the acutely ill patient. Third, the participant must present a summary of the situation and a treatment plan to his supervisor while pursuing management of the patient. For the SID approach, the scenario is stopped at two times: (1) after ordering investigations, and (2) after receiving results of the investigations and prior to the call from the supervisor. The last stop occurs at the end of the scenario.

Based on these principles, the chosen scenario consists of an infant with disseminated herpes simplex virus infection presenting with secondary septic shock. A newborn will present with tachycardia and will evolve towards hypoxemia and hypotension, requiring intubation and volume expansion. Laboratory findings will reveal viral sepsis with leucopenia, thrombocytopenia, increased C reactive protein, and elevated liver enzymes. Finally, skin blisters will appear during the transfer of information from the participant to the supervisor and will confirm the diagnosis.

Figure 3. Clinical reasoning assessment tool is a useful tool for detecting diagnostic errors (such as nonfault, human factors, cognitive, and system-related) and clinical reasoning difficulties according to Kuhn classification (data gathering, data integration, and data confirmation). Instructor has to read questions for each category of error or difficulty and compare with the student performance during the simulation session. By checking all sort or errors concerning clinical reasoning, this tool permit to build specific questions for the student (without feedback for SID approach) or to construct his debriefing (with feedback for classical approach of simulation).



Data Collection and Measurement Tools

Presimulation Questionnaire

This questionnaire will include demographic data (gender, age, year of graduation, number of previous experiences with simulation, learning style, and curriculum followed), degree of self-assessed subjective stress, and self-evaluation of clinical reasoning performances (both using a 10 point Likert-type scale question).

In-Simulation Clinical Reasoning Assessment

In constructing the simulation scenario and vignette, defined cues will be embedded to stimulate hypothesis generation using System-1 of clinical reasoning. After reading the initial patient presentation in the vignette, residents are asked to submit their diagnostic hypothesis by writing. Then, during the scenario, the participant's verbalization (which is also stimulated by given cues from the nurse) of diagnosis coming to mind will be audio recorded. Both the written and audio-recorded hypotheses will be compared to the diagnoses generated by an expert panel that will be submitted to the same scenario. For example, the presence of thrombocytopenia during the data confirmation part should lead to verbalization of (1) bacterial infection, (2) viral infection, (3) intrauterine grown retardation, and (4) platelets immunization. The presence of skin lesions during the data confirmation part should lead to verbalization of (1) herpetic infection, (2) bacterial infection, and (3) varicella infection.

Iterative discussions supported by the Clinical Reasoning Assessment Tool during SID have been designed to allow development and exploration of System-2 of clinical reasoning regarding data collection, diagnostic hypotheses generation, new data interpretation, and management plan. In the classical approach, it is hypothesized that System-2 is discussed during the debriefing period after the simulation. Exploration of how both simulation approaches impact on performance of System-2 will be done during the semistructured interviews (see below).

Postsimulation Questionnaire

Postsimulation questionnaire (Multimedia Appendix 1) will assess residents' self-reported improvement in clinical reasoning and level of satisfaction regarding the simulation approach (both using a 10 point Likert-type scale question). The questionnaire will be designed based on previous literature [63-65] and will be pilot tested with three residents. According to the simulation approach, residents will also complete 5 to 12 questions asking

them to rate statements using a four-point Likert-type scale, ranging from 1 (strongly disagree) to 4 (strongly agree).

Semistructured Interviews

An individual semistructured interview will explore "how" and "why" students' clinical reasoning ability develops through both simulation approaches (SID or classical). Interviews will be conducted using techniques inspired from the explicitation interview during which the interviewer supports the participant, without induction, toward the evocation of a specified experience [66]. After icebreakers, interviews will be constituted of two distinctive parts. First, the interviewer will explore the residents' reasoning process during the simulation experience. Second, he will focus on residents' perception of the simulation experience, including possible advantages and challenges of each simulation type, possible improvements to each methodology, and the perceived impact of SID on the participants' learning. Interviews will be audio recorded and then transcribed for analysis.

Data Analysis

This is an exploratory study that will aid in planning a future larger randomized controlled trial. The target population of residents enrolled in the General Pediatrics and Neonatal-Perinatal Medicine programs represents approximately 50 residents. Based on literature from qualitative research inquiry, the adequate sample size consists of the number of participants at which saturation of data is achieved. This well-described process permits cessation of recruitment when additional data does not bring new properties to unsaturated categories [67]. Different authors agree that saturation is reached after 20 to 25 participants [67,68].

Quantitative data will be analyzed using SPSS 20.0 (IBM SPSS, Chicago, IL). Data will be analyzed using descriptive statistics for all variables. For each Likert-scale type question, a significant cut-off will be pre-established. Comparison of positive versus negative responses will be done with the use of chi-square and Fischer's exact test for nonparametric variables. Number, nature, and order of diagnostic hypotheses will be compared to the responses of a panel of 10 neonatologists from our hospital. A multivariate analysis will be used to investigate potential effects of graduation year, gender, and prior simulation experience on the residents' evaluation of their clinical reasoning. Statistical significance will be defined as a probability value of $<.05$.

Qualitative data will help describe if SID allows better assessment of System-1 and 2 of clinical reasoning compared to the classical approach of simulation. Data from audio-recorded semistructured interviews will be analyzed using NVivo 9.0 software. Analysis will occur as data collection pursues. Recurring themes or distinctive aspects about each student's response will be noted. These notes will be reviewed and expanded as the research continues until saturation of data. Data will be de-identified so that all participants will remain anonymous. Through content analysis, the information will then be categorized according to the principle of convergence [69]. The research team will use deductive analysis and review of all written transcripts.

Results

This study is in its preliminary stages and the results are expected to be made available by April, 2016.

Discussion

Clinical reasoning is an essential skill for everyday medical practice. However, many questions remain regarding how efficiency of the reasoning processes can be most accurately measured [16]. We believe that medical simulation could represent an effective environment for clinical reasoning assessment, as participants are immersed in an authentic and controlled setting [70]. Moreover, SID, an innovative approach to simulation, could provide a closer assessment of both System-1 and 2 of clinical reasoning by the combination of concurrent verbalization and iterative discussions at key steps of the reasoning process.

In this exploratory randomized study, comparing SID to the classical approach of simulation, we expect to find a higher progression of residents' self-assessed clinical reasoning process with SID. More precisely, iterative discussions, by allowing reflection, will lead to improvement in System-2 clinical reasoning process while concurrent verbalization during management of the mannequin will enhance performance of System-1. Verbalization during the classical approach will also have a similar impact on System-1 clinical reasoning. Finally, residents will demonstrate a higher level of satisfaction in the SID approach of SBE.

This is the first randomized study comparing a new simulation approach to the classical mode for developing clinical reasoning skills. In addition, incorporating a qualitative piece to the study with the goal of exploring how each simulation approach impacts on residents' clinical reasoning process is of great interest. Residents of different training levels are included in the study allowing to explore the phenomenon from perspectives of individuals with various levels of clinical reasoning performances. There are also a few limitations to the study. The small sample size does not allow for statistical generalizability of quantitative results. The absence of a robust pre and post assessment of residents' clinical reasoning abilities might not portray the exact impact of each intervention. However, in the absence of a gold-standard tool in clinical reasoning evaluation, this exploratory study could provide detailed and useful preliminary data for the effectiveness of such a new simulation approach. Finally, the case specificity of one unique scenario could limit generalizability of the results.

The findings of the study will be of benefit to medical educators, training programs, and residents who participate in these programs. This study will further our understanding of the complexity of clinical reasoning, and how delivery of the curriculum should be modified to assist residents in better developing their clinical reasoning abilities in neonatology but also in others specialties. This study demonstrates the feasibility of a SBE session where scenarios are built according to clinical reasoning and reflective practice theories and help to put emphasis on clinical reasoning teaching and assessment. For

medical residents, development of a simulation approach that assesses their clinical reasoning abilities will provide them with an opportunity to receive feedback about components of clinical reasoning which need to be improved. It will also provide medical teachers with an opportunity to better understand the students' diagnostic process. Overall, insight into the clinical reasoning process by SBE may contribute to changes in medical education curriculum development and implementation. This may provide residents with better opportunities to develop clinical reasoning by SBE and become clinically competent doctors.

The future should concentrate on optimizing clinical reasoning assessment. A SID session could integrate a combination of well-described clinical reasoning evaluation tools such as script concordance tests [71] or clinical reasoning problems [72]. The validation of such an instrument could provide an essential educational tool for formative or summative assessment of medical students, whatever their level of training or their specialty. In a greater future, assessment of long-term retention of acquired clinical reasoning skills after exposition to SID should be explored once the basics have been settled.

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Conflicts of Interest

None Declared.

Multimedia Appendix 1

Post-simulation questionnaire.

[JPG File, 107KB - resprot_v5i1e26_app1.jpg]

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Abbreviations

NICU: neonatal intensive care unit
PGY: postgraduate year
SBE: simulation-based education
SID: simulation with iterative discussions

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Protocol

Benefits of E-Cigarettes Among Heavy Smokers Undergoing a Lung Cancer Screening Program: Randomized Controlled Trial Protocol

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Abstract

Background: Smoking is a global public health problem. For this reason, experts have called smoking dependence a global epidemic. Over the past 5 years, sales of electronic cigarettes, or e-cigarettes, have been growing strongly in many countries. Yet there is only partial evidence that e-cigarettes are beneficial for smoking cessation. In particular, although it has been proven that nicotine replacement devices may help individuals stop smoking and tolerate withdrawal symptoms, e-cigarettes' power to increase the quitting success rate is still limited, ranging from 5% to 20% dependent on smokers' baseline conditions as shown by a recent Cochrane review. Consequently, it is urgent to know if e-cigarettes may have a higher success rate than other nicotine replacement methods and under what conditions. Furthermore, the effects of the therapeutic setting and the relationship between individual characteristics and the success rate have not been tested. This protocol is particularly innovative, because it aims to test the effectiveness of electronic devices in a screening program (the COSMOS II lung cancer prevention program at the European Institute of Oncology), where tobacco reduction is needed to lower individuals' lung cancer risks.

Objective: This protocol was designed with the primary aim of investigating the role of tobacco-free cigarettes in helping smokers improve lung health and either quit smoking or reduce their tobacco consumption. In particular, we aim to investigate the impact of a 3-month e-cigarettes program to reduce smoking-related respiratory symptoms (eg, dry cough, shortness of breath, mouth irritation, and phlegm) through reduced consumption of tobacco cigarettes. Furthermore, we evaluate the behavioral and psychological (eg, well-being, mood, and quality of life) effects of the treatment.

Methods: This is a prospective, randomized, placebo-controlled, double-blind, three-parallel group study. The study is organized as a nested randomized controlled study with 3 branches: a nicotine e-cigarettes group, a nicotine-free e-cigarettes group, and a control group. The study is nested in a screening program for early lung cancer detection in heavy smokers.

Results: The study is open and is still recruiting.

Conclusions: Stopping or reducing tobacco consumption should be a main goal of any health organization. However, traditional antismoking programs are expensive and not always effective. Therefore, favoring a partial or complete shift to e-cigarettes in

heavy smokers (eg, persons at high risk for a number of diseases) could be considered a moral imperative. However, before following this path, sound and reliable data on large samples and in a variety of contexts are required.

Trial Registration: Clinicaltrials.gov NCT02422914; https://clinicaltrials.gov/ct2/show/NCT02422914 (Archived by WebCite at http://www.webcitation.org/6etwz1bPL)

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KEYWORDS

tobacco cessation; electronic cigarettes; lung cancer screening; smoking related diseases.

Introduction

The World Health Organization estimates that cigarette smoking will claim the lives of 500 million people who are alive today and as many as 1 billion people during the 21st century. Although clinical therapies for smoking cessation have proven effective, the long-term abstinence rate remains low.

Electronic cigarettes, which are also known as tobacco-free cigarettes or e-cigarettes, are battery-operated devices that vaporize a liquid solution of propylene glycol and/or vegetable glycerin in which nicotine and/or flavors are dissolved. A recent review of the field showed that e-cigarettes may be considered safe, with few adverse effects and limited toxicity [1].

The value of these tools is that they reduce the risk of smoking-related diseases. However, while the use of e-cigarettes in heavy smokers will reduce the risk of tobacco-related cancers, their role in antismoking programs has not yet been approved. The World Health Organization and the US Food and Drug Administration have promoted the launch of research on this field of study, but study results are not convergent.

In a prospective study, e-cigarettes were shown to substantially decrease the consumption of tobacco cigarettes without causing significant side effects [2]. In the study, reductions in the number of cigarettes smoked per day and breath carbon monoxide (CO) levels were observed at each visit in all study groups, with no consistent differences among them. Furthermore, rapid improvement in breathing symptoms was observed.

However, most participants continued smoking or started smoking again; after 1 year, fewer than 10% remained abstinent. This is probably due to the research targeting smokers who did not intend to quit. As suggested by Remo and colleagues [3], we argue that much better results might be achieved in smokers motivated to quit.

From a physiological point of view, e-cigarettes appear to eliminate the craving for tobacco in the same way as nicotine replacement therapy (NRT). In an overview of the Cochrane Library [4] that considered studies globally—including more than 50,000 smokers—NRT was described as being particularly efficacious for the short term (3 months) but less so in the long term (12 months). However, the use of NRT increases the success rate of quitting attempts independent of the setting if compared to attempts made by smokers on their own or supported only by counseling. Furthermore, NRT is particularly useful for smokers who are prepared to quit, but who have high nicotine dependence. Eventually, NRT will be particularly effective when smokers' baseline conditions are predictors of successful quitting. Comparing the conclusion of the Cochrane review with the results of studies on e-cigarettes effects, it is clear that in some cases the sample used was quite different from the one used in most trials on NRT. We argue that it is necessary to study e-cigarettes efficacy while also considering population baseline characteristics as well as psycho-cognitive parameters. Indeed, we can expect smokers who are not prepared to quit or in psychosocial conditions associated with a low success rate (ie, mood disturbance, living in a context with high prevalence of tobacco cigarettes smokers, low-income status) to report a worse outcome in studies using e-cigarettes as tobacco cessation treatment [2,4].

Previous research has stressed that monitoring lifestyle parameters (in particular, physical activity and sleep quality) and acting on them could help maintain abstinence. In particular, physical exercise may aid smokers in the first 3 months, while longer effects are less clear [5]. It has been observed that regular physical activity among smokers reduces nicotine withdrawal symptoms and craving. Last, smoking during the night is an indicator of nicotine dependence and predicts failure in smoking cessation [6]. Sleep disturbances have several negative psychological effects, including reduced quality of life and psychological distress (ie, anxiety and depression). Low sleep quality due to abstinence is a predictor of a poor smoking treatment outcome [7].

Low-cost, noninvasive devices are now available to monitor lifestyle parameters. These electronic bracelets are reliable and easy to use. Counseling approaches based on similar tools have been shown to improve outcomes [8].

This protocol may also address a number of psychological parameters, in order to determine whether individual features might hamper behavioral changes and related positive effects on health. Indeed, heavy smokers have specific psycho-cognitive traits [9]. In particular, they generally show higher levels of impulsiveness than nonsmokers. At the same time, smokers tend to have a high level of activity in the behavioral activation system (BAS). It has been suggested that high BAS sensitivity is involved in addictive behaviors like smoking. Individuals with high BAS are more inclined to enact approaching behaviors and experience positive effects when they receive positive rewards.

This study offers an opportunity to test the effectiveness of e-cigarettes in a clinically controlled setting, in order to reduce tobacco consumption and improve health benefits. Furthermore, the protocol is nested in a screening program for early detection of lung cancer at the European Institute of Oncology (IEO) called COSMOS II (Continuous Observation of SMOking

Subjects) that will allow subject recruitment and continuous monitoring. The COSMOS II project aims to improve early diagnosis of lung cancer, which is currently considered to be the most important life-saving tool. This is an Italian program, coordinated by IEO and created to identify an optimal personalized protocol for early diagnosis in people with a high risk of lung cancer (ie, heavy smokers or former smokers over the age of 55). The COSMOS II program will enroll 10,000 heavy smokers or former smokers throughout Italy. COSMOS II derives from the previous successful COSMOS I screening project [10,11].

The main hypothesis is based on previous research on the effect of e-cigarette use in substitution of tobacco cigarettes. We hypothesize that the reduction of cigarette tobacco consumption leads to a significant decrease of cough, breath shortness, and other respiratory symptoms at 6 months. This reduction will improve the quality of life and well-being. Furthermore, we expect this effect to be higher for smokers using nicotine e-cigarettes and support than for a placebo group and support-only group. This would be coherent with previous research on NRT and e-cigarettes. Indeed, we argue that smokers who are both motivated to start a quitting attempt and aware of smoking-related risks yet continue consuming several tobacco cigarettes a day need an integrated antismoking strategy that combines physiological, behavioral, and psychological interventions. The use of electronic cigarettes, providing both physiological and behavioral replacement of tobacco cigarette consumption, and low-intensity counseling, providing psychological support, might then be an optimal strategy in these cases.

Eventually, we expect particular psychological conditions to be associated with better outcomes. In particular, we hypothesize that participants with a low level of depression, an active lifestyle, and a low BAS will find the use of e-cigarettes more advantageous.

Methods

Objectives

The main objective was to evaluate the impact of a 3-month e-cigarettes program to reduce smoking-related respiratory symptoms (eg, dry cough, breath shortness, mouth irritation, and phlegm) as a consequence of reduced tobacco cigarette consumption. Secondary objectives were to (1) assess the success rate of smoking cessation attempts in the three groups; (2) monitor safety and toxicity during the study; (3) evaluate psychological and behavioral (ie, lifestyle) effects of e-cigarettes; (4) assess the impact of e-cigarette use on quality of life; and (5) identify cognitive/behavioral patterns as e-cigarettes success predictors in reducing tobacco cigarette smoking.

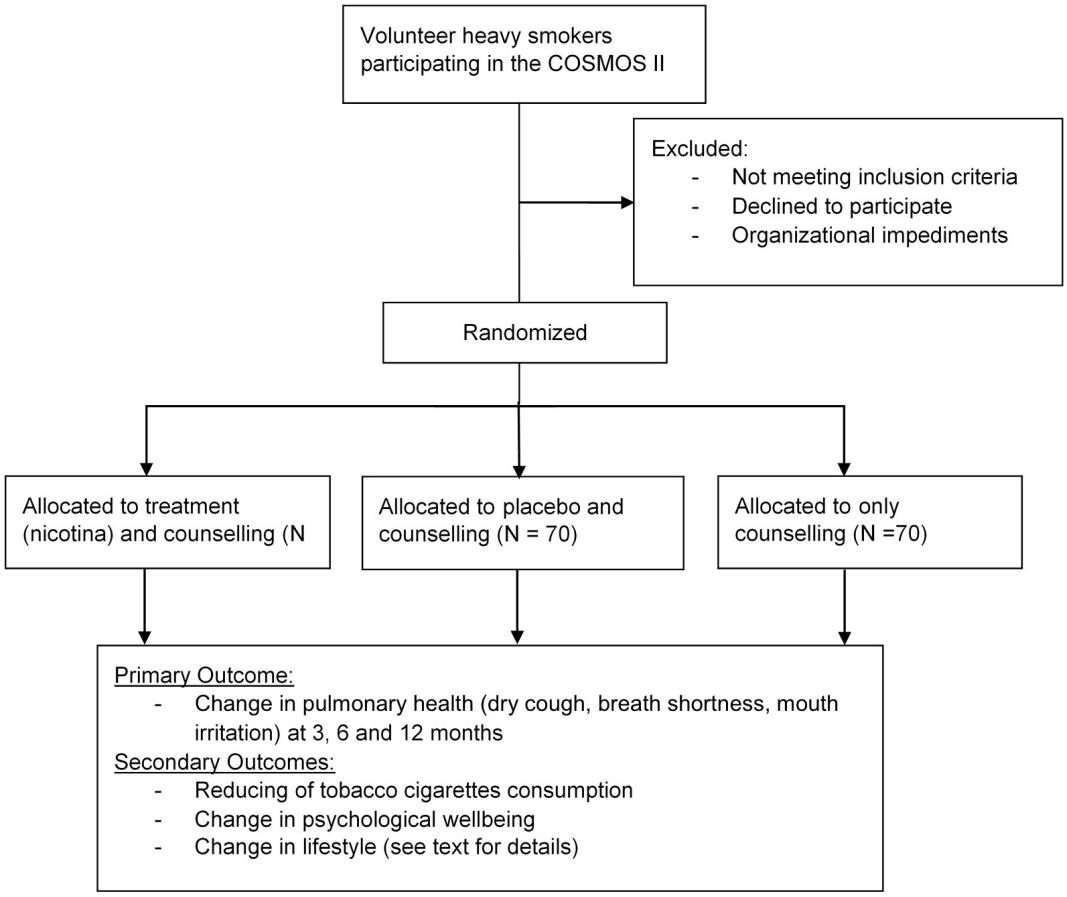
More specifically, the main aim of the project concerns the effectiveness of e-cigarettes in improving lung health in the heavy smokers involved in the COSMOS II program. If proven safe and effective, e-cigarettes should be included in lung cancer screening programs as a standard tool to reduce smoking-related risks for lung diseases. Naturally, this aim requires a scientific approach, since e-cigarettes should not increase nicotine dependence. Another fundamental aim of the project regards the effectiveness of e-cigarettes in reducing tobacco consumption. In particular, no studies to date have tested the feasibility and effectiveness of these tools in limiting risky behaviors (eg, tobacco smoking) among heavy smokers enrolled in a lung cancer screening program. Consequently, we want to determine whether providing e-cigarettes to participants in a controlled protocol reduces tobacco consumption, as well as related health and breathing problems. We also aim to analyze the psychological characteristics and needs of the subjects enrolled in the COSMOS II program, in order to evaluate how risk perception (eg, the premise of risky behavior adoption) is associated with a psycho-cognitive profile. We argue that an important and successful screening project, such as COSMOS, should incorporate a comprehensive approach to the individual.

Design

This is a prospective, randomized, placebo-controlled, double-blind, three-parallel group study. The study protocol was designed using the recommendations of the Consolidated Standards of Reporting Trials statement (Figure 1).

For this study, we opted for the VP5 electronic cigarettes kit, which offered a good quality/price ratio and proven reliability and safety. Nicotine and nicotine-free liquids are produced by BioFumo, which fully collaborated with us and provided liquids in nicotine concentrations of 8 mg/mL and packages that were not distributed for commercial use.

Figure 1. CONSORT flow diagram.



Participants

Volunteer smokers are recruited from among COSMOS II participants at the IEO hospital. Details on the inclusion and exclusion criteria are provided in the "Selection Criteria" section below.

The main inclusion criterion is adult healthy smokers who voluntarily choose to take part in a lung cancer screening program. This screening program includes both a low-dose computed tomography (CT) scan and blood tests in order to detect early signs of lung cancer. Consequently, all of our participants agree to undergo these examinations and are over the age of 55 at the beginning of the study. To be included in the study, COSMOS II participants must have smoked an average of 10 cigarettes or more a day for at least the past 10 years. Furthermore, they also must report strong motivation to stop smoking as measured by a motivational questionnaire (see the "Instruments and Measures" section).

Since we are interested in assessing the effect of a specific e-cigarettes-based treatment, we exclude smokers already using e-cigarettes, which we define as smokers who had ever regularly used e-cigarettes for more than 1 week alone or in combination with tobacco cigarettes. Thus, all participants are inexperienced with the use of e-cigarettes (full instructions are provided by the researcher in charge of the study during the briefing). Also, smokers who at the moment of the interview are undergoing NRT or underwent NRT in the previous 6 months are excluded. In this way, we tried to prevent any psychological and physiological confounding effects due to previous treatments. Furthermore, people with a history of psychiatric, severe dyspnea, and cardiovascular diseases are also excluded.

All the including and excluding criteria are evaluated during the first clinical examination of the COSMOS II program. Only after the clinical examination, which includes objective tests and an anamnestic interview by a physician in charge, is a smoker considered for possible inclusion in the protocol.

Selection Criteria

Inclusion Criteria

1. Subjects are involved in the COSMOS II study
2. Subjects have smoked at least ten cigarettes a day for the past 10 years
3. Subjects wish to reduce tobacco smoking (motivational score higher than 10) who are not treated at a smoking center
4. Signed informed consent

Exclusion Criteria

1. Symptomatic cardiovascular disease
2. Symptomatic severe respiratory disease
3. Regular psychotropic medication use
4. Current or past history of alcohol abuse
5. Use of smokeless tobacco or NRT
6. Participation in another antismoking program in the current year

Treatments

Group 1 Treatment (E-Cigarette and Support)

The participants receive an e-cigarettes kit and 12 10-mL liquid cartridges containing an 8 mg/mL concentration of nicotine. They are instructed to use the electronic cigarette ad libitum during the first week before their quitting day (determined at the first contact) in order to familiarize themselves with its use. Starting at Week 2 (soon after the designated quitting day), the participants are asked to stop smoking tobacco cigarettes and use the e-cigarettes exclusively for the next 11 weeks. E-cigarette use is monitored through a weekly paper diary and regular telephone interviews. Since it is not possible to exclude tobacco-cigarette smoking after the quit day, the weekly diary also contains items related to cigarette smoking. Tobacco smoking is also addressed during the periodic calls.

During treatment, a low-intensity remote (by phone) counseling program is provided to maintain motivation and monitor any psychological and/or physical problems related to the study protocols. This program includes 4 calls in total, 1 at the ends of Week 1, Week 4, Week 8, and Week 12. Each call will last about 10 minutes; will address concerns about any ongoing psychological, physical, and behavioral changes; and will support the participants' continued motivation by providing practical suggestions.

The participants also receive an electronic fitness bracelet in order to assess their physical activity and sleep quality during treatment.

Group 2 Treatment (Placebo)

The participants receive an e-cigarettes kit and 12 10-mL nicotine-free liquid cartridges. This liquid has the same manufacturer, flavor, and components (except for nicotine) as the one used in Group 1. All other procedures and measures used for Group 2 are the same as those for Group 1.

Group 3 Treatment (Control/Support-Only)

The participants are provided only with low-intensity remote (by phone) antismoking counseling to motivate and support cigarette smoking cessation and abstinence. Scheduling, aims, and structure are the same as those of Group 1. The participants also receive an electronic fitness bracelet in order to assess their physical activity and sleep quality during treatment. All measures and procedures are identical to Groups 1 and 2.

Treatments Common to All Groups

- A complete explanation of the project and informed participant consent
- Baseline behavioral, motivational, and psycho-cognitive evaluation (set of questionnaires)
- Baseline clinical parameter assessment
- Initial and final face-to-face interview
- Regular telephone interviews
- Explanation of the weekly short-diary procedure
- Briefing on and delivery of the e-bracelet
- Low-intensity remote smoking cessation counseling program

Instruments and Measures

Clinical Parameter Evaluation

- Anamnesis
- Clinical examination
- CO measurement
- Low-dose CT scan
- Circulating micro-RNA examination
- Respiratory examination

Instruments

- Self-reported measures are used for cough and other respiratory symptoms assessment. We opted for Likert scales to measure cough, breath shortness, mouth irritation, and phlegm frequency as well as the Leicester Cough Questionnaire.
- Fagerstrom Test for Nicotine Dependence: a 6-item self-reporting questionnaire assessing nicotine dependence. It requires a few minutes to complete [12]. This test is administered to all COSMOS II participants.
- Motivational questionnaire [13]: a 4-item self-reporting questionnaire assessing motivation to quit smoking. The total classifies the patient into 1 of 4 motivational categories (from "not ready to quit" to "highly motivated"). This test is administered to all COSMOS II participants.
- Hospital Anxiety and Depression Scale (HAD): The HAD is a self-administered questionnaire composed of two 7-item scales, 1 for anxiety and 1 for depression, which should be used as 2 separate measures of emotional distress. The scale has been validated for Italian culture by Costantini and showed high internal consistency with Crohnbach alpha, ranging from .83 to .85 [14]. The HAD evaluates symptoms of anxiety and depression, avoiding misattribution due to the physical aspects of the illness. The values range from 0 to 21 for each scale. Cutoff scores are preliminarily defined as normal (0-5), mild (6-8), moderate (9-11), and severe (greater than 11) for both anxiety and depression patients [15].
- BIS/BAS Scale [16]: a 20-item self-reporting questionnaire evaluating the behavioral inhibition system (BIS) and BAS. Each of the items is rated on a 5-point scale, ranging from 1 (does not describe me at all) to 5 (describes me very well).
- Barratt Scale [17]: an 11-item self-reporting questionnaire designed to measure impulsiveness. All items are measured on a 4-point scale (Rarely/Never; Occasionally; Often; Almost Always/Always), where a 4 generally indicates the most impulsive response, although some items are scored in reverse order to avoid a response bias [18].
- The Leicester Cough Questionnaire: a valid, self-reported cough-specific health status measure. It is a 19-item questionnaire that has been validated in acute and chronic cough [19]. The overall score ranges from 3 to 21 with a higher score indicating a better quality of life.
- Electronic bracelet (the Flex FitBit): a device that allows lifestyle monitoring of physical activity and sleep characteristics, including: sleeping and napping (hours slept, light vs deep sleep, and waking periods); activity (distance, calories burned, activity time, and activity intensity); nutrition (food and beverage intake); mood (assessment of

the affect state); and an insight engine that identifies hidden connections and patterns in day-to-day activities.
- The exhaled CO is measured by the The Micro+ Smokerlyzer, which has less than 5% H_2 cross-sensitivity.
- Ad hoc questionnaire: measures demographic data, self-perceived quality of life (analogue scale), physical activity, and smoking-related issues (characteristics of the smoking experience).

Recruitment and Follow-Up

All inclusion and exclusion criteria are checked during the registration procedure and initial assessment for inclusion in COSMOS II. Eligible participants are asked to provide informed consent. The informed consent form is signed and dated by both the participant and the physician. Enrolled participants receive a chronological number and are assigned to a treatment group (e-cigarettes with nicotine, e-cigarettes without nicotine, or control).

A randomization list using a permuted block design (40 blocks of 6 subjects randomly assigned to 1 of the 3 treatment groups) have been previously prepared by an independent personnel unit and labeled with the progressive number applied to the packaging containing e-cigarettes and liquid cartridges with or without nicotine (Group 1 and Group 2).

Neither the participant nor the researcher in charge knows whether the liquid in the e-cigarettes kit contains nicotine. Only the statistician who prepared the randomization list and the person labeling the e-cigarettes packaging know the actual treatment.

Each participant is then assigned to one of the three groups and receives the related treatment, as illustrated above.

Follow-up: 6 months (at the IEO)

- Behavioral psycho-cognitive questionnaires
- Clinical parameters assessment (respiratory symptoms)
- Smoking status assessment (questionnaire and CO level). The nonsmoking status is established by self-report items and the CO level (ppm<5).
- Debriefing, during which we also ask participants of Group 1 and Group 2 to guess if they used a nicotine-free or a nicotine-based e-cigarette.

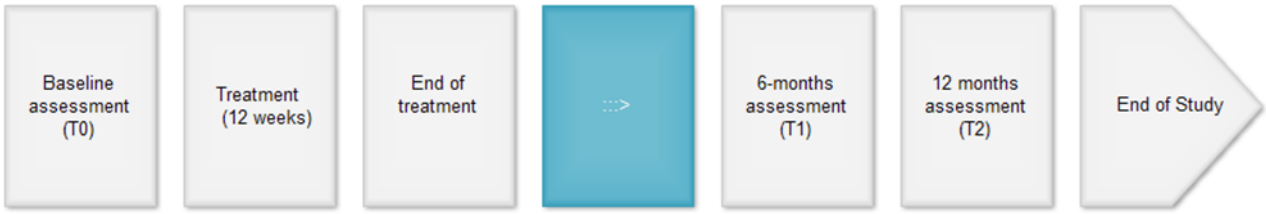
End-point: 12 months (at the IEO, during the annual assessment of COSMOS II)

- Clinical parameter assessment (respiratory symptoms)
- Final behavioral and psycho-cognitive assessment
- Smoking status assessment (questionnaire and CO level)
- Debriefing and collection of comments

Timeline (Figure 2):

- Distribution of e-cigarettes: 3 months (ends at Week 12)
- Data tracking (e-bracelet) and active monitoring: 6 months (ends at Week 24)
- End point: 12 months

Figure 2. Protocol plan.



Endpoints/Evaluation Criteria

Primary Outcome Measures

- Change in respiratory symptoms (eg, dry cough, breath shortness, mouth irritation)

To evaluate the impact of a 3-month e-cigarettes program to reduce smoking-related respiratory symptoms (dry cough, breath shortness, mouth irritation, and phlegm) through reduced tobacco cigarette consumption. The primary outcome is measured at Month 6 and then at the follow-up at Month 12.

Secondary Outcome Measures

- Change in psychological well-being (HAD scale)
- Change in number of cigarettes smoked daily
- Change in the concentration of exhaled air CO
- Change in daily activity (mean number of daily steps)
- Change in lifestyle as measured by ad-hoc questionnaires

Statistical Considerations

Sample Size

Starting with data provided by previous studies on the effect of smoking discontinuation, whether using or not using e-cigarettes [2,20,21], we expected to find a reduction between 20% and 30% of respiratory symptoms reported by participants. We used cough as the measure to power the trial on. Using a two-sided *Z* test, a sample of 70 participants in either of the experimental groups (e-cigarettes with or without nicotine) and 70 in the control group (counseling alone) will reach 80% power, at .05 significance level, to detect a 20% reduction in the frequency of symptoms from the baseline in either of the e-cigarette groups (with or without nicotine) compared to a 5% reduction in the control group (counseling alone).

Statistical Plan

The main analysis will consist of comparing the reduction in symptoms after 6 months for the three groups: e-cigarettes with nicotine vs control; e-cigarettes without nicotine vs control; and

e-cigarettes with nicotine versus e-cigarettes without nicotine. Analyses will be based on two-sided *Z* tests.

A secondary analysis will assess the reduction of the consumption of tobacco-containing cigarettes after 6 months among the groups. Analysis will be based on paired Student's t-test or the Wilcoxon signed rank tests.

No interim monitoring is planned since, given the sample size, any interim analysis would have too few events to be interpreted. Efforts will be made to maximize retention by maximizing trust at the baseline assessment and collecting multiple means to contact and commit participants, including email and phone contact details, and by assertive follow-up. Starting with our previous experience with a similar population coming from the COSMOS I program, we expect that our participants will be compliant with and committed to the aims of the study. Giving the expected intrinsic motivation of participants and thanks to the above strategies, the study aims to have at least 80% retention at 6 months and 70% at 12 months. Considering these figures, we expect to maintain a statistical power to detect a reduction of 5 cigarettes/day in our smokers (the cigarettes per day mean is about 20 in the COSMOS population). Thus, using a two-sided two-sample *t*-test with a significance level (alpha) of .05, a sample size of 49 participants per arm will achieve 80% power to detect a mean reduction of 5 cigarettes/day between any of the 2 experimental arms and the control arm, assuming a mean consumption of 20 cigarettes/day in the control arm and a common standard deviation of 8.7.

Ethical Considerations

This study is performed in accordance with the principles stated in the Declaration of Helsinki and subsequent amendments, and in accordance with the Good Clinical Practice Guideline. This protocol was assessed and certified by the ethical board of Fondazione Umberto Veronesi, the ethical committee of Università degli studi di Milano, and the ethical committee of the European Institute of Oncology. The ClinicalTrials.gov identifier is NCT02422914. Informed consent will be obtained from all subjects.

Results

At the time of manuscript submission, the trial's status is "recruiting."

Discussion

Principal Findings

Cigarette smoking is a major risk factor for a variety of diseases. The World Health Organization states that tobacco kills nearly 6 million people each year and that an annual death toll of more than 8 million is expected by 2030.

Despite the availability of approved medications and smoking cessation aids (ie, NRT, bupropion, varenicline, and counseling programs), long-term quitting rates are relatively low. Most smokers try to quit without professional help even when they see their doctor on a regular basis. Indeed, smoking status is rarely documented and smoking cessation treatments are offered even less frequently [22], wasting an opportunity provided by the doctor-patient relationship.

The failure of tobacco control is clear in developing countries. However, in most rich countries, tobacco control also is problematic due to a number of factors. Economic and ethical issues often conflict in antismoking research and strategies [23], and the relatively recent availability on the market of e-cigarettes has added further confusion [24]. Consequently, independent studies are needed in this field to avoid any possible external influence. Furthermore, we believe that lung cancer screening programs (like COSMOS II, where this project is nested) have an ethical obligation to provide participants access to all the information and strategies that could help them to reduce their risk. Stopping or reducing tobacco consumption, then, should be a primary goal. However, traditional antismoking programs are expensive and not always effective. Therefore, favoring a partial or complete shift to e-cigarettes in heavy smokers (eg, persons at high risk for a number of diseases) could be considered a moral imperative. However, to follow this path, sound and reliable data is required on a large sample and in a variety of contexts.

Last, the question of the use of e-cigarettes and their regulation concerns not only physiological and toxicity aspects (eg, the association between cancer and smoking), but also certain relevant behavioral and psychological aspects as well. Thus, it is necessary not only to understand the toxicity of these new ways of smoking, but also their impact on smokers' minds and lifestyles.

Conclusions

E-cigarettes-based intervention could provide a gateway to boosting health-related behavior changes, in order to reduce tobacco consumption and positively impact smokers' quality of life. Indeed, reducing tobacco consumption or supporting abstinence could be considered a fundamental aim of a screening program, since a change in smoking habits reduces the risk of smoking-related diseases.

Recent studies [2,3,25] show that e-cigarettes must be considered safe devices that are potentially useful both for reducing clinical symptoms (eg, cough, phlegm, breath shortness) and enhancing the impact of antismoking interventions. Consequently, the use of e-cigarettes is particularly important in prevention programs and for high-risk subjects. Many aspects are currently unclear and debated [25]. Hence, the outcome of this study will provide important data on the possible role of e-cigarettes as tools for use in screening and prevention programs. We argue that e-cigarettes apply a medicalized substitution logic in which nicotine dependence becomes a route to health in addition to a disorder to be treated.

Acknowledgments

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Nicotine and nicotine-free liquids are produced by BioFumo, which offered full collaboration to meet the study demands. In particular, BioFumo provided us with the liquids with nicotine concentration of 8 mg/mL in packages that were not distributed for commercial use. All devices and products (eg, e-cigarettes, liquids, electronic bracelets) are purchased thanks to economic support provided by the FUV. No support is provided to the study by tobacco industries or other for-profit corporations.

We are grateful to IEO personnel for their help in organizing and managing the study, as well as for their contribution to methodological and ethical aspects.

Authors' Contributions

CL, MM, GP, PM, and GV conceived of and designed the study. CL coordinated the study, CL and CP acquired legal authorizations, and MM and SS managed participants. Statistical support and data management were provided by PM and RB, while e-cigarettes and liquids were managed by JC and EOS. Drafting and writing of the manuscript was handled by CL, MM, and PM. All authors have read and approved the final manuscript.

Conflicts of Interest

None declared.

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Abbreviations

BAS: behavioral approach system
BIS: behavioral inhibition system
CT: computed tomography
CO: carbon monoxide
COSMOS: Continuous Observation of SMOking Subjects
HAD: Hospital Anxiety and Depression Scale
IEO: European Institute of Oncology
NRT: nicotine replacement therapy

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Original Paper

A Web- and Mobile-Based Intervention for Women Treated for Breast Cancer to Manage Chronic Pain and Symptoms Related to Lymphedema: Randomized Clinical Trial Rationale and Protocol

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Abstract

Background: Despite current advances in cancer treatment, many breast cancer survivors still face long-term post-operative challenges as a result of suffering from daily pain and other distressing symptoms related to lymphedema, ie, abnormal accumulation of lymph fluid in the ipsilateral upper limb or body. Grounded in research-driven behavioral strategies, The-Optimal-Lymph-Flow is a unique Web- and mobile-based system focusing on self-care strategies to empower, rather than inhibit, how breast cancer survivors manage daily pain and symptoms. It features a set of safe, feasible, and easily-integrated-into-daily-routine exercises to promote lymph flow and drainage, as well as guidance to maintain an optimal body mass index (BMI).

Objective: To conduct a randomized clinical trial (RCT) to evaluate the efficacy of the Web- and mobile-based The-Optimal-Lymph-Flow system for managing chronic pain and symptoms related to lymphedema. The primary outcome includes pain reduction, and the secondary outcomes focus on symptom relief, limb volume difference by infra-red perometer, BMI, and quality of life (QOL) related to pain. We hypothesize that participants in the intervention group will have improved pain and symptom experiences, limb volume difference, body mass index, and QOL.

Methods: A parallel RCT with a control-experimental, pre- and post-test, repeated-measures design is used in this study. A total of 120 patients will be randomized according to the occurrence of pain. Participants will be recruited face-to-face at the point of care during clinical visits. Participants in the intervention group will receive the Web- and mobile-based The-Optimal-Lymph-Flow intervention and will have access to and learn about the program during the first in-person research visit. Participants in the control group will receive the Web- and mobile-based Arm Precaution program and will have access to and learn about the program during the first in-person research visit. Participants will be encouraged to enhance their learning by accessing the program and following the daily exercises during the study period. Participants will have monthly online self-report of pain and symptoms at 4 and 8 weeks post-intervention. During the two in-person research visits prior to and 12 weeks post-intervention, participants will be measured for limb volume difference, BMI, and complete self-report of pain, symptoms, self-care behaviors, and QOL.

Results: This trial is currently open for recruitment. The anticipated completion date for the study is July 2017. The primary endpoint for the study is absence or reduction of pain reported by the participants at week 12 post-intervention.

Conclusions: The-Optimal-Lymph-Flow is a unique Web- and mobile-based self-care and patient-reported outcome system designed to effectively help women treated for breast cancer manage daily pain and symptoms related to lymphedema. Patients learn self-care strategies from a Web- and mobile-based program and track their symptoms. The RCT will directly benefit all women treated for breast cancer who suffer from or at risk for pain and symptoms related to lymph fluid accumulation.

Trial Registration: Clinicaltrials.gov NCT02462226; https://clinicaltrials.gov/ct2/show/NCT02462226 (Archived by WebCite at http://www.webcitation.org/6du4IupG5)

(JMIR Res Protoc 2016;5(1):e7) doi:10.2196/resprot.5104

KEYWORDS

pain; ache; soreness; tenderness; symptoms; lymphedema; breast cancer; health behavior; mHealth

Introduction

Background

Annually, more than 230,000 women are diagnosed with breast cancer, and currently there are more than 2.9 million breast cancer survivors in the United States [1]. Even years after cancer treatment, about 20-40% of women treated for breast cancer suffer daily from chronic pain and more than 50% of women report multiple distressing symptoms related to lymphedema (ie, the abnormal accumulation of lymph fluid in the ipsilateral upper limb or body) [2-5]. The abnormal accumulation of lymph fluid or lymphedema after breast cancer treatment is a result of obstruction or disruption of the lymphatic system associated with cancer treatment (eg, removal of lymph nodes and/or radiotherapy), influenced by patient personal factors (eg, obesity or higher body mass index [BMI]), and triggered by factors such as infections or trauma [6-8].

Breast cancer survivors without a diagnosis of lymphedema also suffer from pain (40%), tenderness (47.3%), aching (30%), or soreness (32.7%); however, significantly higher number of breast cancer survivors with lymphedema experience pain (45.2%), tenderness (52.4%), aching (61.9%), or soreness (31%) in the ipsilateral upper limb or body [9]. In addition to pain, on average, breast cancer survivors without lymphedema report about 5 distressing symptoms while breast cancer survivors with lymphedema report 10 distressing symptoms related to the accumulation of lymph fluid [9-10]. It is clear that many breast cancer survivors still face long-term post-operative challenges as a result of suffering from daily pain and other distressing symptoms related to lymphedema, despite current advances in cancer treatment.

Pain and symptoms related to the accumulation of lymph fluid following breast cancer treatment remain as the main debilitating late complications that impact the breast cancer survivors' quality of life [2,3,5,11]. Persistent pain related to cancer treatment is considered a stressful complication since it is perceived as a constant reminder of cancer [2,12] and exerts tremendous limitations on breast cancer survivors' daily living [2,5]. Pain and other distressing symptoms related to lymphedema following cancer treatment can instigate fears and induce feelings of loss of control [2,3,5]. Specifically, the experience of pain, including tenderness, aching, or soreness, causes significant and unrelenting distress among breast cancer survivors [3]. Such distress is usually heightened when breast cancer survivors expect pain and symptoms related to lymphedema to disappear but instead stay as a "perpetual discomfort" [3]. The negative impact of pain and symptoms related to lymphedema can be a source of considerable disability and psychological distress that negatively influences the patient's daily living [2,3,11,12], and creates a tremendous burden on the health care system [13]. Nonetheless, in clinical practice pain and symptoms related to lymphedema are still under-recognized and undertreated.

While more research is needed to explore the exact etiology of persistent pain and other symptoms after breast cancer treatment,(eg, arm swelling, breast swelling, chest wall swelling, heaviness, firmness, tightness, stiffness, numbness, burning, stabbing, tingling, and limited limb movement), physiologically, the accumulation of lymph fluid in the affected area or limb may create undue pressure on nerves, producing feelings of pain, aching, tenderness, soreness, burning, tingling, stabbing, and numbness as well as inducing sensations of swelling, heaviness, tightness, and firmness [14-15]. Accumulated lymph fluid in the affected area or limb also leads to stiffness and limited limb movement of arm, shoulder, fingers, and elbow [10,15]. Significant associations are found between pain (including aching and tenderness) and accumulation of lymph fluid in the ipsilateral upper limb [10,15]. Research has also shown that with increased number of symptoms reported, breast cancer survivors' limb volume increased [10,15]. Limb volume as detected by the infra-red perometer has significantly elevated as breast cancer survivors' reports of pain, tenderness, aching, swelling, heaviness, firmness, and tightness have increased [10]. On average, breast cancer survivors reported 4 symptoms for those with <5.0% limb volume increase; 5 symptoms for 5.0-9.9% limb volume increase, 7 symptoms for 10.0-14.9% limb volume increase, and 13 symptoms for >15% limb volume increase, respectively (P<.001) [10].

Breast cancer survivors are known to have a compromised lymphatic system due to breast surgery, dissection of lymph nodes and vessels, and radiation, which leads to ineffective lymphatic drainage, thus accumulated lymph fluid in the affected area or limb [10,15,16]. In addition to the risk factor of compromised lymphatic drainage from cancer treatment, higher BMI is also an established risk factor for the accumulation of lymph fluid [6-10]. Physiologically, a larger body mass creates a disproportion in lymph transport and capacity, resulting in excess extracellular fluid [6,17]. Women are 1.11 times more at risk for developing lymphedema with every increase of 1kg/m^2 in their BMI [6-8,16]. Although the known risk factors for symptoms related to accumulation of lymph fluid directly

from cancer treatment cannot be avoided, (such as removal of lymph nodes, surgery, radiation, chemotherapy, and hormonal therapy), some risk factors, (such as compromised lymphatic drainage and higher BMI), can be modified through education and self-care strategies [14,18,19].

Patient education focusing on self-care strategies holds great promise for reducing the risk of lymph fluid accumulation [14,18,19]. Research evidence demonstrates that even after controlling for confounding cancer treatment-related risk factors, patient education on self-care strategies remains an important predictor for patient-centered outcomes, including symptom experience and self-care behaviors [14,18,19]. Risk factors, such as compromised lymphatic drainage and higher BMI, can also be modified through self-care strategies [14,19]. Current patient education emphasizes precautionary lifestyle behaviors, such as avoidance of repetitive limb movement, lifting weighted objects, needle punctures, blood draw, and the use of compression garments for air travel in the affected limb [20,21]. To date, there is a paucity of high quality evidence to support these precautionary practices that reduce the risk of lymphedema and relieve pain or symptoms related to lymph fluid accumulation [20,21]. Research is lacking to provide evidence to reduce pain and symptoms related to lymph fluid accumulation through self-care strategies targeting compromised lymphatic drainage and higher BMI.

Grounded in research-driven self-care behavioral strategies [14,19], The-Optimal-Lymph-Flow [22] is a unique patient-centered Web- and mobile-based educational and behavioral program focusing on self-care strategies to lessen the symptom burden by promoting lymph flow and maintaining optimal BMI, targeting compromised lymphatic system and BMI, that is, risk factors for pain and symptoms related to lymph fluid accumulations. Patients learn self-care stratigies through the Web- and mobile-based program which can be downloaded on computer, laptop, as well as any mobile phones and tablets. Its underlying premise is to empower, rather than inhibit, how breast cancer survivors live their lives by emphasizing “what to do,” rather than “what to avoid.” It features a safe, feasible, and easily-integrated-into-daily-routine self-care strategies that include shoulder mobility exercises to promote shoulder function, muscle-tightening breathing, muscle-tightening pumping exercises, and large muscle exercises to promote lymph flow and drainage, as well as general instructions to encourage nutrition-balanced (more vegetables and fruits), portion-appropriate diet (feeling 75% full for each meal), adequate hydration, and sleep to strive for maintaining optimal BMI. Patients can learn and follow all the exercises through avatar video simulations [14,19]. The efficacy of The-Optimal-Lymph-Flow has been demonstrated in our recently published study of 140 patients who received the face-to-face nurse-delivered program [19]. Findings of the study demonstrated that over 90% of patients improved their limb volume at 12-month follow-up. This system has been used successfully for its usability testing. The preliminary usability tested was completed by 30 breast cancer survivors who evaluated the easiness, difficulties, and feasibility of using the system on computer, iPhone, iPad, or other smartphones or tablets. Findings of the usability and feasibility test have demonstrated that patients love the Web-based program, especially the videos using the avatar technology to demonstrate the complicated lymphatic system and illustrate the physiological functions of each exercise and detailed step-by-step instructions for each exercise.

The purpose of the research is to conduct a randomized clinical trial (RCT) to evaluate the efficacy of the Web- and mobile-based The-Optimal-Lymph-Flow system, a patient-centered educational and behavorial symptom management program focusing on promoting lymph flow and optimizing BMI, for managing chronic pain and symptoms related to lymphedema.

Objectives and Hypotheses

The primary objective of this study is to determine the effectiveness of the Web- and mobile-based The-Optimal-Lymph-Flow system for managing chronic pain, aching, soreness, and tenderness among breast cancer survivors and quality of life related to pain. We hypothesize that participants in the intervention group will have no or less severe pain, aching, soreness, and tenderness, and better quality of life related to pain, aching, soreness, and tenderness in comparison with participants in the control group.

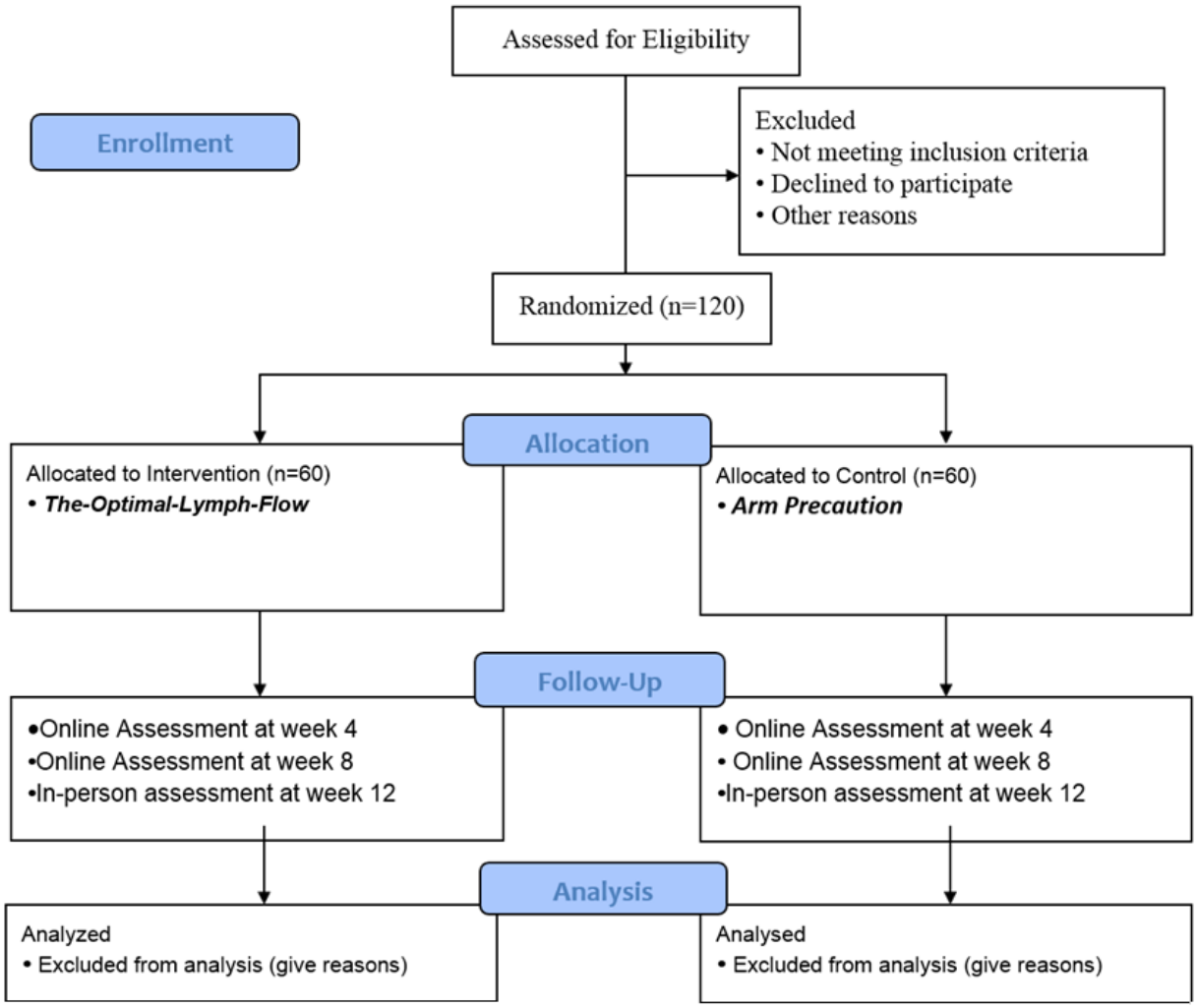
The secondary aim of the study is to evaluate the effectiveness of the Web- and mobile-basedThe-Optimal-Lymph-Flow system for managing symptoms related to lymph fluid accumulation, limb volume differences, and BMI. We hypothesize that participants in the intervention group will have fewer or less severe symptoms related to lymph fluid accumulation, minimal limb volume differences, and better BMI in comparison with participants in the control group.

Methods

Design

For this project, chronic pain, including aching, tenderness, soreness, is defined as persistent or intermittent pain in the ipsilateral upper limb or body for more than 3 months after surgical treatment for breast cancer, that is, beyond the expected period of healing [21-23]. A 12-week, two-arm, parallel randomized controlled trial (Clinical trial registration ID: NCT02462226) has been designed to evaluate the effectiveness of the Web- and mobile-based The-Optimal Lymph-Flow self-care strategies to promote lymph flow versus control Arm Precaution group for managing chronic pain and symptoms related to lymphedema. The data collectors will be blinded to the group assignments. The protocol is in accordance with the CONSORT-EHEALTH (see Multimedia Appendix 1) checklist [24]. Multimedia Appendix 2 presents the full proposal review feedback form. Figure 1 shows the CONSORT-EHEALTH flow diagram for recruitment and randomization [24].

Figure 1. The CONSORT flow diagram.



Ethical Approval

This study was approved by the Institutional Review Board of NYU Langone Medical Center on June 8, 2015.

Study Population

Study population includes: (1) patients who have been surgically treated for breast cancer more than 3 months (healing usually occurs within 3 months of surgical treatment for cancer); (2) patients who report *persistent or intermittent* pain, including aching, tenderness, soreness; (3) patients may or may not report any of the symptoms related to lymphedema (ie, swelling, heaviness, tightness, firmness, numbness, tingling, stiffness, limb fatigue, limb weakness, and impaired limb mobility of shoulder, arm, elbow, wrist, and fingers); (4) patients may or may not have a history of lymphedema or have been treated for lymphedema; (5) patients have Internet access to the Web- and mobile-based program at home or willing to access the program using the laptop provided by the researchers at the cancer center; (6) ability to understand and the willingness to sign a written informed consent document.

Exclusion criteria are (1) patients who do not report any pain, including aching, tenderness, and soreness; (2) patients who have known metastatic disease or other bulk disease in the thoracic or cervical regions; (3) patients who have lymphedema due to cancer recurrence; (4) patients with documented advanced cardiac or renal disease.

Recruitment

Recruitment Process

Participants have been recruited face-to-face at point of care during clinical visits from New York University (NYU) Cancer Center. To accomplish recruitment of 120 participants, we plan to use the successful procedures of recruiting and consenting participants used by the PI and the team in the preliminary studies [3,9,14,17,19]. Successful strategies include the use of *Invitation Flyer* that describes the study. This *Invitation Flyer* is posted on the bulletin boards or breast cancer support website at the cancer center, and is also available in the reception areas of the cancer center, examination rooms, and rooms holding support group meetings. In addition, health care providers such as nurses, oncologists, breast surgeons, and oncology radiologists at the center are willing to refer women meeting the inclusion criteria to the study by distributing the *Invitation Flyer* that describes the study to the potential participants.

Consent Process

After reading the *Invitation Flyer*, if a woman is interested in participating in the study, she would schedule a meeting with the research coordinator at that time or at other convenient time for them. During the meeting, the research coordinator will confirm her interest, determine if the woman is eligible for the study and the research coordinator will again explain the study in detail and provide enough time for the woman to ask questions. If the woman agrees to participate, she will sign the consent form.

Confidentiality

Confidentiality will be maintained. Patients will be assigned a study ID specific for the study. Study data recording for the research will only use the study ID without the patients' identifying information. A document file that has the patients' study ID with the patients' identifying information will be separately stored in locked files accessible only to the research coordinator, research nurse, or the PI. Electronic data will be stored in a password protected computer accessible only to the PI, the research nurse, and the research coordinator. Data analysis will be carried out and reported in the aggregate data so that individual identities are not revealed. Careful training and supervision of research coordinator and research nurse will insure study procedures are carried out in accordance with established protocols.

Randomization and Blinding

The randomization assignment will be generated by our senior statistician using a computer-generated randomization procedure. Participants will be randomized based on their report of pain/aching/soreness or tenderness to be allocated to intervention and control group. The researchers who perform pre- and post-intervention measurements will be blinded throughout the study to the participants' assigned arm. Participants will not know which intervention was the intervention of interest and which one was the comparator.

Study Intervention

Overview

The Web- and mobile-based *The-Optimal-Lymph-Flow* [19,22] includes information about lymphedema, diagnosis and measurement of lymphedema, lymphatic system, risk of lymphedema, self-care, daily exercises, arm precautions, and ask experts. Participants in the intervention group will have access to the 8 Avatar videos that provide step-by-step instructions for The-Optimal-Lymph-Flow exercises to promote lymph flow and optimize shoulder and limb mobility. The platform also has a section entitled Arm Precautions, representing current patient education that emphasizes precautionary lifestyle behaviors, such as avoidance of repetitive limb movement, lifting weighted objects, needle punctures, blood draw, and the use of compression garments for air travel in the affected limb [20,21]. Figures 2-5 shows some screenshots of the The-Optimal-Lymph-Flow program. Table 1 presents the strategies, rationales, and actions for *The-Optimal-Lymph-Flow* program.

Table 1. The-Optimal-Lymph-Flow program: self-care strategies, rationales, and actions

Strategies	Rationales	Actions
Promoting lymph flow		
Muscle-tightening deep breathing	The whole body lymph fluid has to be drained through the lymphatic ducts above the heart. Muscle-tightening deep breathing stimulates lymphatic ducts and help lymph fluid drain.	At least twice a day in the morning & at night before brushing teeth or as much as the patient wants throughout the day.
	Lymph fluid drains when muscles move. Muscle-tightening deep breathing creates the whole body muscle movements that create muscle milking and pumping action and help to drain lymph fluid.	Air-travel: before take-off and after landing. Sedentary lifestyle: at least every 4 hours.
Muscle-tightening pumping	Muscle-tightening pumping exercises create arm muscle pumping. This helps lymph fluid flow and decreases the fluid build-up in the arms.	At least twice a day in the morning & at night before brushing teeth or as much as the patient wants throughout the day.
	Muscle-tightening pumping exercises build the arm muscle that helps lymph fluid flow and drain.	Air-travel: before take-off and after landing. Sedentary lifestyle: at least every 4 hours.
Large muscle exercises: walking, marching at home, dancing, swimming, Yoga, Tai Chi	Large muscle exercises create muscle milking and pumping to promote overall body lymph fluid flow and drain.	At least 10-minutes daily. Air-travel: get up and walk around for flight over 4 hours. Sedentary lifestyle: get up and walk at least every 4 hours.
Improving limb functional status		
Shoulder exercises	Improved limb mobility after surgery facilitates local muscle movements that create muscle milking and pumping to promote local limb lymph fluid flow and drain.	One week after surgery if there is no surgical drains or after the surgical drains are removed. At least twice a day until limb functions are returned to normal. Whenever limb mobility is limited throughout the recovery.
Keep a healthy weight		
Eat nutrition-balanced diet (ie, more vegetables and fruit as well as quality proteins); Maintain portion-appropriate diet (feeling 75% full for each meal)	Overweight or obesity is an important risk factor for lymph fluid accumulation. Having extra weight makes it difficult for lymph flow and drain. This can lead to extra lymph fluid build-up. There are numerous weight management programs available to assist with weight loss. Although there are a lot of weight reduction programs, each person may respond differently to each program. The core of the weight management is to eat a nutrition-balanced, portion-appropriate diet. It is also important to stay hydrated, exercise, and get adequate sleep.	Each meal daily It is important to talk to the nutritionist who can help to find proper weight reduction programs.
Stay hydrated	People may actually be thirsty, not hungry.	Drink 6 to 8 glasses of water daily; in the morning, before and during meals, and throughout the day. Avoid drinks with calories (eg, juices). Drink green tea to boost metabolism.
Large muscle exercises	Daily large muscle exercises (eg, walking, running, swimming, Yoga) help to burn more calories. Daily large muscle exercises also promote lymph flow by creating muscle pumps.	At least 30-minutes 3 times a week or daily.
Get enough sleep	Lack of sleep increases the production of the stress hormone cortisol, creates hunger, and leads to overeating. Getting just one more hour of sleep per night reduces belly fat accumulation.	At least 7-8 hours of sleep per night.

Figure 2. The Web- and mobile-based The-Optimal Lymph-Flow.

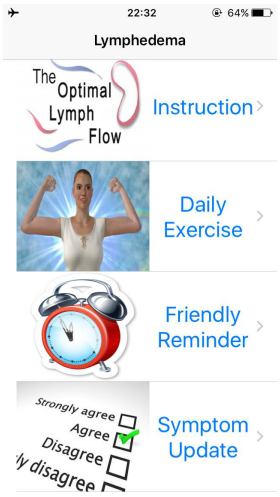


Figure 3. Video instructions for daily exercises.

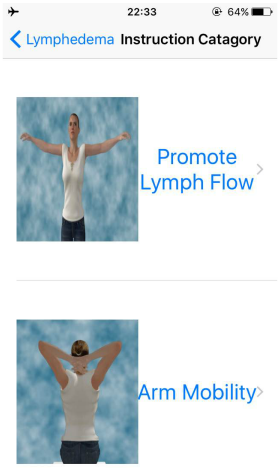


Figure 4. Symptom reporting.

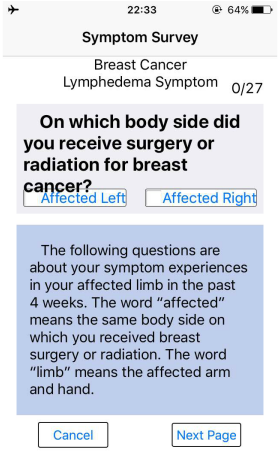
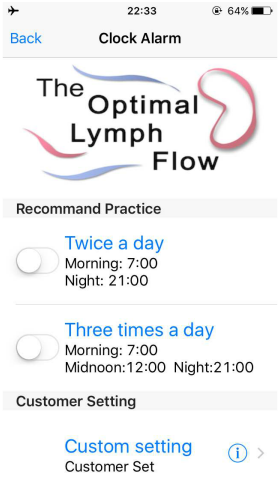


Figure 5. Friendly reminder for daily exercises.



The-Optimal-Lymph-Flow Intervention Group (n=60)

Patients assigned to The-Optimal-Lymph-Flow intervention group will have access to the Web- and mobile-based The-Optimal-Lymph-Flow platform and they will learn about theprogram and daily exercises during the first in-person research visit. Patients will have access to the website contents of Lymphedema, Diagnosis of Lymphedema, Lymphatic System, Self-care, Daily Exercises, and Ask Experts. Patients will also have access to the 8 avatar videos that provide step-by-step instructions of daily exercises to promote lymph flow and optimize shoulder and limb mobility. In addition, the patients will be introduced to an app, and have the choice to use either the Web-based program or the app for daily exercises. However, patients *will not* have access to the section Arm Precautions since the participants in the intervention group will receive comparable information as in the Arm Precautions section, but with particular emphasis on "what to do," rather than "what to avoid."

Control Arm Precaution Group (n=60)

Patients assigned to the control Arm Precaution group will have access to the website section that emphasizes on precautionary lifestyle behaviors, such as avoidance of repetitive limb movement, lifting weighted objects, needle punctures, blood draw, and the use of compression garments for air travel in the affected limb [20,21]. Patients will have access to the following contents of the website: Lymphedema, Diagnosis of Lymphedema, Risk of Lymphedema, Lymphatic System, 3 avatar videos for Daily Exercises to promote limb mobility, and Arm Precautions. However, patients will not have access to The-Optimal-Lymph-Flow program, including Self-care, Daily Exercises to promote lymph flow, and 8 Avatar videos as well as Ask Experts that allows them to email to the researchers about their self-care. Patients will access to the Web- and mobile-based Arm Precaution program and learn about the program and daily exercises to promote limb mobility during the first in-person research visit.

Duration of Intervention

From the previous one-arm clinical trial and usability and feasibility studies on the Web- and mobile-based The-Optimal-Lymph-Flow [19,23], we estimate that it would take about 45-60 minutes for patients to learn all the sections of the program and about 15 minutes to learn the The-Optimal-Lymph-Flow exercises for the intervention group through 8 avatar videos. It takes about 5 minutes to perform a set of The-Optimal-Lymph-Flow daily exercises each time. We encourage patients in the intervention group to perform at least twice a day or more times of the exercises during the study period. Participants in the control group will have access to three limb mobility exercise avatar videos and it takes less than 3 minutes to perform a set of limb mobility exercises each time. Patients in the control group will be instructed to perform limb mobility exercises at least twice a day or more times during the study period.

Data Collection

Data Collection Procedures

Data will be collected at baseline prior to intervention, and at week 12 post-intervention. Data collection at each time point will take approximately 30 minutes. Within one week of enrollment for the clinical trial, patients will have baseline assessment of pain and symptoms, limb volume difference, BMI, and quality of life. The follow-up assessment will occur at week 12 post intervention.

Two In-Person Research Visits: (1) prior to intervention there will be baseline assessment of pain and symptoms, limb volume difference, BMI, and quality of life; and (2) 12 weeks post-intervention assessment of pain and symptoms, limb volume difference, BMI, self-care behaviors, and quality of life.

Two Online Assessments: patients in the intervention and control group will receive an email that provides a link to assess pain and symptoms as well as quality of life at week 4 and week 8 post-intervention. Confidentiality of the patients will be protected for the online assessment since patients will use their study ID to access the online assessment.

Outcome Measures

Demographic and Medical Information: a structured tool is used to gather demographic and medical information and is verified through reviewing participants' medical records [14,17,19]. The demographic and medical information will be considered as covariates, including expectation of the program,

pain medications, age, surgeries, lymph nodes procedure, radiation, chemotherapy, time since surgery, time since lymphedema diagnosis, and hormonal therapy.

Primary and Secondary Outcome Measures: primary measure focuses on pain which is assessed during prior to the intervention and week 12 post-intervention in-person visit, as well as week 4 and 8 post-intervention online assessment. Secondary measures include symptoms, limb volume difference by infra-red perometer, BMI, quality of life related to pain. Limb volume difference by infra-red perometer and BMI are only measured prior to and week 12 post-intervention in-person visits. Symptoms and quality of life related to pain are assessed prior to and week 12 post-intervention in-person visit as well as week 4 and 8 post-intervention online assessment.

Pain and Symptoms Related to Lymphedema: the Lymphedema and Breast Cancer Symptom Experience Index is a valid and reliable self-report tool to assess pain, including aching, soreness, tenderness, as well as symptoms related to lymphedema (ie, arm swelling, breast swelling, chest wall swelling, heaviness, firmness, tightness, stiffness, burning, stabbing numbness, tenderness, stiffness, redness, blistering, and tingling (pins and needles) [14,17,19]. Each symptom can be treated as categorical variable by choosing a “Yes” or “No” to indicate the presence or absence of a given symptom. Each item can also be rated on a Likert-type scale from 0 (no presence of a given symptom) to 4 (greatest severity of a given symptom). Higher scores indicate more severe symptom presence. A response frame of last three months will be used for all participants to ensure the chronicity of symptom presence during the first in-person visit.

Limb Volume Difference by Infra-Red Perometer: perometry 350S will be performed on each arm as it is held horizontally. The perometer maps a 3-dimensional graph of the affected and non-affected extremities using numerous rectilinear light beams, and interfaces with a computer for data analysis and storage. A 3-dimensional limb image will be generated and limb volume will be calculated. This optoelectronic method has a standard deviation of 8.9 ml (arm), less than 0.5% of limb volume with repeated measuring [17,19] .

Quality of life Related to Pain: the Pain Impact Questionnaire (PIQ-6), a reliable and valid six question health survey, will be used to measure pain severity and the impact of pain on an individual's functional health and well-being. The PIQ-6 measures the severity of pain and its impact on work and leisure activities, as well as on emotional well-being within a variety of diseases and general populations. High PIQ-6 *T* scores indicate greater pain impact/worse health [21-23].

Height, Body Weight, and BMI: height will be measured to the nearest 0.1 cm with a portable stadiometer (Scale-Tronix 5002 Stand on Scale, Scale-Tronix Company, Carol Stream, IL, USA) without shoes [25]. An electrical device (InBody 520, Biospace Co, Ltd, Seoul, Korea) will be used to measure the participants’ body weight, BMI is calculated using the formula: weight (kg)/height (m^2) [25].

Practice of Self-Care Behaviors: Risk Reduction Behavior Checklist is a structured self-report checklist that will be used to quantitatively and qualitatively assess patients’ practice of self-care behaviors at the study endpoint of 12-week after intervention [17,19]. The checklist include a list of self-care behaviors that promote lymph flow, eg, muscle-tightening deep breathing, muscle-tightening pumping, shoulder exercises, large muscle exercises, and having nutrition-balance and portion-appropriate diet, adequate hydration and sleep, as well as compression therapy for lymphedema.

Statistical Analysis

Primary Endpoint

The primary endpoint for the study is absence of pain or pain reduction reported by the participants at week 12 post-intervention.

Sample Size and Power Calculations

We will enroll a total of 120 participants: 60 participants in The-Optimal-Lymph-Flow intervention and 60 participants in the Arm Precaution control group to account for a potential attrition of 20%, which has been observed in the prior studies in breast cancer survivors [10]. This will yield an adequate analytic sample size. Even with 20% attrition based on a 2 sample 2-sided *t* test with alpha=.05 and power of 90%, we can detect a difference of 0.7 standard deviations in the difference between the presence of pain in the intervention group compared to the control group at 8 weeks or at 12 weeks. The projected sample size will also provide sufficient statistical power for mixed regression models. For linear mixed models of continuous outcomes (eg, pain ratings), statistical power will exceed 80% to detect a medium effect, assuming a constant group effect, correlations of r=.5 between observations, alpha=.05, and compound symmetry of the covariance structure.

For binary outcomes, based on the three repeated observations with a conservative estimate for the assumed correlation of r=0.5 between observations, alpha=.05, sample size of n=50 per treatment arm will have power of 80% to detect odds ratios of the difference between groups ranging from 2.6 to 3.5 (small to medium effects).

Adequacy of sample size will be monitored during the analyses in three ways: (1) assessing sufficiency to estimate the number of model parameters identified by preliminary analyses; (2) examining model fit indices (eg, intra-class correlation coefficient to assess fit of random effects) to ensure data adequately support models generated, and (3) assessing the adequacy to accurately estimate model parameters by generating confidence intervals.

Analysis Plan

We will summarize graphically and numerically the distributions of pain, aching, soreness, tenderness, other symptoms related to lymph fluid accumulation, limb volume differences, BMI, and quality of life as well as covariates (such as medication for pain, age, or education and treatment variable and limb volume and self-care behaviors). The proportion of individuals experiencing pain, aching, soreness, tenderness will be compared between the intervention and control group over time, as will the mean severity ratings and associated quality of life scores. Mean comparisons of number of reported symptoms, limb

volume, and BMI will be conducted between the intervention and control group as well as over time.

Bivariate relationships between the variables and treatment group will be assessed using point-biserial correlations and phi coefficients to identify potential covariates for inclusion into statistical models. Linear mixed effects models will be used to analyze continuous outcomes (eg, ratings pain, aching, soreness, tenderness, symptom ratings, and quality of life) and generalized linear mixed models will be used to analyze binary outcomes (presence of pain, aching, soreness, tenderness, and total scores of quality of life). These models will incorporate fixed effects for time, group, and any identified covariates. As indicated by preliminary models to estimate variance effects, models will include random effects for subject-specific slopes and intercepts. A statistically significant fixed effect for group by time interaction will indicate a treatment effect, with the direction of difference determined from mean values. Models will be compared for goodness of fit and modifications to link functions, distributional form, and correlation structure will be made as necessary.

Interim Analyses

No interim analysis will be conducted since the intervention is only 12-week long.

Method of Handling Missing Data and Non-Adherence to Protocol

Data from participants who are missing >20% of any scale will be excluded from calculation of that scale, though remaining data meeting requirements for completeness will be retained. Analysis of missing data will first determine whether it can be assumed to be missing at random (MAR) or not missing at random (NMAR). Based on the results of this step, appropriate methods will be selected for addressing missing data (eg, Heckman Selection for NMAR, Multiple Imputation for data which are MAR).

Further, mixed effects regression models are robust in the presence of missing data. Unlike traditional repeated measures designs (eg, repeated measures ANOVA) which employ listwise deletion, excluding all records for individuals who miss a single observation, mixed effects models are capable of incorporating all completed observations for estimations. Although our previous studies suggest that we will have a limited number of missed observations, individuals need not have the same number of observations. This maximizes the statistical power of our analyses and reduces the likelihood of systematic bias in estimates.

Results

This trial is currently open for recruitment. The anticipated completion date for the study is July 2017. The primary endpoint for the study is absence or reduction of pain reported by the participants at week 12 post-intervention.

Discussion

Hypothesis

Low cost and pragmatic self-care strategies for symptom management may hold great promise for improving patients' quality of life [6,19]. To date, limited research has been designed to help breast cancer survivors to manage their daily distressful symptoms, including pain. This clinical trial focuses on primary outcomes of pain reduction and secondary outcomes of relief of symptoms related to lymphedema, limb volume difference by infra-red perometer, BMI, quality of life to refine procedures and estimate effect size for the future efficacy of multi-center RCT. We hypothesize that participants in the intervention group will experience no or less severe pain, aching, soreness, and tenderness and better associated quality of life (related to pain, aching, soreness, and tenderness) in comparison with participants in the control group when compared to participants in the control Arm Precaution group.

Conclusions

The proposed project will directly benefit all women treated for breast cancer who suffer from or are at risk for pain and symptoms related to lymph fluid accumulation by providing a low-cost, technologically-driven delivery model to universally expand the accessibility of The-Optimal Lymph-Flow. With health care reform under way, using Web- and mobile-based technology to develop low cost and pragmatic patient-centered intervention is the key to lessening the health care cost and advancing the science of symptom management.

Acknowledgments

The study, entitled "*The-Optimal-Lymph-Flow*: An e-Health Approach to Enhancing Management of Chronic Pain and Symptoms Related to Lymphedema among Women Treated for Breast Cancer," is supported by Pfizer Independent Grants for Learning & Change (IGL&C) (grant #13371953) and Judges and Lawyers Breast Cancer Alert (JALBCA) with Mei R Fu as the principal investigator.

Conflicts of Interest

None declared.

Multimedia Appendix 1

CONSORT-EHEALTH checklist V1.6.1.

[PDF File (Adobe PDF File), 558KB - medinform_v5i1e7_app1.pdf]

Multimedia Appendix 2

Full proposal review feedback form.

[PDF File (Adobe PDF File), 332KB - medinform_v5i1e7_app2.pdf]

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Abbreviations

BMI: body mass index
MAR: missing at random
NMAR: not missing at random
PIQ-6: Pain Impact Questionnaire-6
RCT: randomized controlled trial

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Protocol

Development, Validation, and Evaluation of Web-Based Iranian Diabetic Personal Health Record: Rationale for and Protocol of a Randomized Controlled Trial

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Abstract

Background: Diabetes is one of the four main types of noncommunicable or chronic diseases. Iran is among the countries with the highest incidence of diabetic patients. A study demonstrated that the collection of diabetic data is neither organized nor standardized. There is currently no instance of electronic personal health records particularly used for diabetic patients in Iran, hence the need for one, which will be useful for self-care of diabetic patients.

Objective: The objective of the study is to examine the impact of a Web-based diabetic personal health record (DPHR) on the self-care status of diabetic patients as compared with the control group.

Methods: This study is a randomized control trial, which involves a systematic review of literature of the preferred data elements regarding a DPHR, and reevaluating the results with the opinions of local endocrinologists. Inclusion criteria were as follows: type 2 diabetic patients between 20-70 of age who live in the Mashhad City and having the disease for at least one year. The sample size is 72 people that were randomly assigned to the control and intervention groups. The participants in the intervention group were allowed access to the Web-based DPHR system, while those in the control group will continue to receive the usual care for 4 months. The study primary outcome measures include self-care status of participants and planned visit adherence.

Results: At the moment, there is an ongoing recruitment of participants, and preliminary results will be published in early 2016.

Conclusions: We expect the final DPHR model, developed and tested during this study, to help diabetic patients to actively participate in their care management process, and also to empower the physician in providing more quality informed decisions regarding their patients.

Trial Registration: irct.ir IRCT2013082914522N1; http://www.irct.ir/searchresult.php?id=14522&number=1 (Archived by WebCite at http://www.webcitation.org/6cC4PCcau).

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KEYWORDS

diabetes mellitus; type 2; personal health record; web-based; Iran

Introduction

Diabetes Definition

Diabetes is one of the four main types of chronic or noncommunicable diseases [1], and its management is of great concern to the society and world at large [2-5]. A definition by the World Health Organization [6] states that: "Diabetes is a chronic disease that occurs either from insufficient production of insulin by the pancreas or ineffective use of produced insulin". Type 2 diabetes results from the body's ineffective use of insulin.

Lack of or insufficient care for a diabetic patient can result in several complications such as diabetic neuropathy, retinopathy, nephropathy, diabetic foot, and myocardial infarction [2]. However, continuous blood glucose and blood pressure monitoring, timely visits, appropriate physical activity, blood lipids control, and periodical examinations, as foundation of diabetes self-management, can help patients lessen these complications [7-9].

In recent times, there has been increasing prevalence of diabetes globally, with the low- and middle-income countries having the highest prevalence. The global prevalence of diabetes was estimated to be 9% in 2014 [10], with an estimated increase of 366,000,000 worldwide and 6,421,000 in Iran by 2030 [11,12]. In order to manage the increasing number of diabetics in the future [13,14], and to reduce the workload of health care providers, there is a need to redefine the role of diabetes management centers [15]. Patients who have more knowledge about their disease and procedures are more proficient at communicating experiences and then become a useful asset in long-term care [16]. In this way, Web-based personal health records (PHRs) provide patients with access to their health information [17-19].

Personal Health Records

Application of PHRs, in general, and diabetes-specific PHRs, has been growing very rapidly. Studies have demonstrated that Web-based diabetes management tools can improve self-care activities [19] and biomedical outcomes measures such as glycated hemoglobin (HbA1c), low-density lipoprotein (LDL) cholesterol, blood pressure, and body mass index (BMI) [20-24]. The benefits of PHRs in supporting self-management is clear, especially in facilitating communications among health care providers and supporting information access [25].

Iran is among the countries with the highest incidence of diabetic patients [26]. A recent study demonstrated that the collection of diabetic data are neither organized nor standardized [27]. There are no diabetic PHRs (DPHRs) used to organize diabetes data in the country.

This multiphase study involves: (1) a systematic review of literature regarding the preferred data elements regarding a DPHR, (2) an afterward refinement of data elements by the local endocrinologists, (3) a systemic development of a Web-based DPHR application (app), and (4) final evaluation of the model through a randomized controlled trial. The researchers hope that the final model developed throughout in this study will help diabetes patients to actively participate in their treatment plan, and also optimize the decision making process of the diabetes physicians in their everyday practice.

Study Hypothesis

The participants assigned to receive the DPHR tool will manage better self-care as compared to those who receive only usual care.

Methods

Randomization and Blinding

After the provision of informed consent, participants are randomly allocated to the control or intervention arm stratified by gender (male, female), employment status (employed and nonemployed), and age groups (=<30; 30-50; and =>50 years of age), that is, the covariate adaptive random allocation is considered. The random allocation sequence will be done through a concealed and computerized random number program [28]. A person who has no direct involvement in this trial will do the randomization process. Also, we note that the participants and attending physician (RA) can't be blinded to the use of PHRs because the artefact is obvious, but the chief researcher (MT) and data analyst (MAA) are blinded to the participants during the trial. To minimize contamination of the trial, the subjects in the intervention arm will be required not to interact and share their DPHRs information with other participants in the control arm.

Study Design

A four-phase approach, including a systematic review of evidence regarding DPHR development, usability testing, iterative refinement, and intervention evaluation will be utilized. A randomized controlled trial (RCT) with a 2-arm parallel design and allocation ratio of 1:1 will be used to evaluate the impact of DPHRs on self-care. Repeated assessments will be conducted at two time points, including baseline and postintervention, over a 4-month trial period. The patients in the intervention group will be allowed access to the Web-based app, while those in the control group will be receiving usual care throughout the study period.

The DPHR interface will be designed in three types, covering our three groups of users including the participating patients, the clinic staff, and the physicians. The chief researcher will be the system administrator. Reminders via a short message service (SMS), telephone contact, and email will be used for visiting the website.

Trial Population and Recruitment Procedure

The inclusion criteria for participants are as follows: the patient must be a male or female with type 2 diabetes, age between 20 and 70 years, with a minimum diabetes history of one year (according to the primary diagnosis date), must live in the city

of Mashhad, must be fairly computer literate, and have access to the Internet. The included participants must be able to provide informed consent.

We excluded the already included participants, if they, for any reason, decided to terminate the participation, or were unable to actively continue the required cooperation, due to sickness, pregnancy, and etc.

The trial will be conducted in Mashhad City (Iran), with participants being recruited from one endocrinology practice unit. The number of diabetic patients is estimated to be above 120,000 in Mashhad [29]. We will start the study with the acquisition of baseline information including age, education level, gender, self-care score, HbA1c, weight, employment status, length of disease, and diagnosis date along with the information required to check the inclusion eligibility. Only the qualified participants will be given the consent form.

After signing the consent form, participants will be randomized into either the intervention or the control arm. After providing confirmed consent, a package including a copy of the consent form, welcome letter, and a take-home manual, along with step-by-step directions for using the website will be rendered manually to the participants. The participants are allowed to communicate with the trial team through email or telephone, sharing their questions and concerns. For assuring the quality of trial, the trial assistant will be trained during several sessions regarding the interaction with the website, the potential questions and answers of the participants, and the intervention process.

Ethical Considerations

The Research Review Committee as well as the Regional Ethics Committee (approval # 921835) approved the trial. The trial is registered on the Iran Registry of Clinical Trials [30].

The informed written consent will be obtained from the eligible participants, with its content clearly explained to the subjects by the trial assistant. During the project, participants will be given means of communication in order to share their questions and concerns.

The research tools, including questionnaires, will be completely anonymous. However, a unique code will be included in each form in order to manage further references. Access to the DPHR Internet app requires a user name and password, which are provided by the trial assistant. All participants' information will be securely stored and will only be accessible to authenticated trial team members.

Intervention Development

The Web-based DPHR app is designed to empower diabetes patients with self-care information and tools. The major aim of this study is to systematically develop an evidence-based Internet app, which is ultimately validated by the local endocrinologists. The app allows participants to easily enter their monitoring data, to view their history of progress, to get informed about their appointment schedule, and to learn about their disease. The patients will also be able to share the blood glucose and lab result trends with their physicians for further advices.

The participants in the intervention arm will receive the Web-based app as well as usual care. On the other hand, the control arm will continue to receive usual care.

The trial team and the software provider will be in charge of the DPHR development, updates, and maintenance. The app interface will be evolved and optimized throughout the trial using heuristic usability evaluation techniques [31] by medical informatics specialists, endocrinologists, and also the participants.

The trial will last for 4 months. The major treatment process delivered by the attending physicians will not have any impact on the intervention and vice versa. During the study, participants may receive extra visits and lab tests in addition to their routine care plan, in order to keep careful track of changes in their health information.

Sample Size

For estimation of sample size, we did not find any study regarding self-care in Iranian population. However, a study has demonstrated that self-care activities and quality of life are correlated [32]. Therefore, sample size for this study was estimated based on the similar study in the country of Iran [33]. A sample size of 60, corresponding to the formula, has been estimated. Considering confidence interval 95%, a power of 80%, and a dropout rate of 20%, 72 subjects will be required. A total of 36 subjects will be randomly assigned to the intervention arm to receive the DPHR tool, and the remaining 36 subjects assigned to the control to receive the usual care.

Outcome Measure

The investigators will employ the Summary of Diabetes Self-care Activities Measure-revised for assessing self-care behaviors [34]. A study has reported the validity and reliability of this instrument, with an internal consistency of 0.47 and a correlation of 0.4 [34]. Also, the patient adherence to a planned visit will be measured.

In this study, researchers will examine any potential correlation, rather than cause and effect, between the app usage and the diabetes follow-up clinical indicators. Such indicators which are based on the routine follow-up procedure of diabetes clinics are: fasting blood sugar (FBS); 2-hour postprandial blood sugar; bedtime blood sugar; blood pressure; weight; height; BMI; lipid profile (total cholesterol, triglyceride, HDL, and LDL); and HbA1c.

Data Collection

One primary outcome and several secondary outcomes will be assessed with several validated assessment tools. Face and content validity of the questionnaire will be assessed through the expert opinions (including two endocrinologists, one health informatics specialist, and one methodologist) and valid literature. Also, the questionnaire will be completed using structured interviews through the person who is not a member of trial team. The subjects will complete the questionnaire on paper.

Each participant will be requested to complete the questionnaire in-person at the reference clinic at the time of the scheduled

diabetes follow-up. Statistics on website visits will be obtained using the access logs by the Web server. After the end of the trial period, the participants' opinions and experiences on the DPHR tool will be assessed using another questionnaire.

Analysis Plan

All analyses will be 2-tailed and statistical significance is considered as a *P* value of ≤.05. Data will be analyzed using SPSS. χ^2 test will be used for determining differences in subject groups based on data collected from baseline, and postintervention (significance interval of 95% will be considered). A *t* test will be used to compare the control and intervention groups, while a chi-square test will be used for continuous and categorical variables, respectively. The distribution of variables, website usage statistics, and website user satisfaction are examined using descriptive analysis.

Examination of the impact of the DPHR tool on self-care will be performed using a linear mixed model at the end of the trial period. The longitudinal nature of data collection will result in a repeat in the measure of analysis of variance using data from all trial assessment points. Missing values will be handled using multiple imputation [35]. All analyses will be done according to intention-to-treat principle.

Before inferential analysis, the homogeneity of variances of distributions in the study groups will be confirmed. The sequential logistic regression will be used for adjusting baseline characteristics and possible confounders. Dropouts from the study will be measured by no-use of the app tool for more than one month. Also, participants in the intervention group must enter blood glucose measurements at home (FBS, bedtime, 2 hours after lunch, and 2 hours after dinner) at least four times a week, otherwise, they are eliminated from the trial. The access data will be assessed using the Web server access log.

Results

The recruitment of participants is still ongoing and preliminary results will be published in early 2016.

Discussion

New Insights for the App

The results from this trial will propose new insights on the app of DPHRs for patient involvement and participation with their care process. Initially in this trial, we will identify the most important data elements to consider in the design of a Web-based tool for improvement of self-care activities related to diabetes patients and then we will assess the effect of this systematically developed tool.

These trial findings will be of great help and usefulness to endocrinologists who are currently facing the challenge of timely and rapid treatment decision making in the ambulatory settings. The results are likely to be generalizable to other ambulatory endocrine/diabetes centers.

It is envisaged that results from this trial could improve diabetes patients in terms of self-care activities. In addition, our results might be used as a basis in further similar research aiming at patient empowerment using Web-based tools.

The strengths of our trial design are the evidence-based development process of the DPHR tool (based on a systematic review), the inclusion of local experts' opinions, and the eventual refinement of the adopted design using the usability techniques. Both the value of evidence-based content development and the importance of usability testing in the app development process have been emphasized in several studies [36-38], pointing to the facts that such considerations have rarely been used across similar works [36].

Limitations

There are a few limitations in our study; these are listed in the following paragraphs.

The primary need to recruit participants with minimum computer and Internet literacy might be a serious limiting factor. Generally, in Internet-based interventions, issues such as digital divide, computer literacy, age, and interest in technology can be effective in participant recruitment. Usually, the younger people, computer literates, and those with Internet access have the most tendency in participating in such studies. This trial is not an exception in principle. Also, a study has shown that middle-age well-educated women who are reasonably well off financially tend to participate in PHR research [37]. Such tendencies may present bias in our findings, and thus our trial may not necessarily represent the actual distribution of the population being studied.

The trial sample will only represent type 2 diabetes patients. It is possible that the findings will not be generalizable to other types of diabetes disease.

Due to the nature of intervention, the investigators may frequently request the presence of the subjects for the interviews. This may cause discomfort and stress to some of the participants. To address such issues, we will provide financial incentives, such as free visits or free laboratory testing in order to encourage better involvement.

Another potential limitation for this trial is the attrition rate. It is possible to have a high rate of participant dropout and subsequently a significant loss of data. Therefore, reminders via telephone contact, email, and SMS will be used.

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Authors' Contributions

MT is the principal investigator and conceived the trial. He was responsible for overall administration of the grant. MT and AA were primarily responsible for development of the DPHR app. ZMK and RA assisted in trial coordination. MAA provided expertise in the RCT design and analysis.

Conflicts of Interest

None declared.

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Abbreviations

app: application
BMI: body mass index
DPHR: diabetic personal health record
FBS: fasting blood sugar
HbA1c: glycated hemoglobin
LDL: low-density lipoprotein
PHRs: personal health records
RCT: randomized controlled trial
SMS: short message service

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Protocol

Primary Care Pathway for Childhood Asthma: Protocol for a Randomized Cluster-Controlled Trial

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Abstract

Background: Asthma is the most common chronic condition in children. For many, the disease is inadequately controlled, which can burden the lives of children and their families as well as the health care system. Improved use of the best available scientific evidence by primary care practitioners could reduce the need for hospital care and improve quality of life and asthma control, thereby reducing overall costs to society and families.

Objective: The Primary Care Pathway for Childhood Asthma aims to improve the management of children with asthma by (1) providing primary care practitioners with an electronic guide (a clinical pathway) incorporated into the patient's electronic medical record, and (2) providing train-the-trainer education to chronic disease management health professionals to promote the provision of asthma education in primary care.

Methods: The research will utilize a pragmatic cluster-controlled design, quantitative and qualitative research methodologies, and economic evaluation to assess the implementation of a pathway and education intervention in primary care. The intervention will be analyzed for effectiveness, and if the results are positive, a strategy will be developed to implement delivery to all primary care practices in Alberta.

Results: The research has been successfully funded and ethics approvals have been obtained. Practice recruitment began fall 2015, and we expect all study-related activities to be concluded by March 2018.

Conclusions: The proposed pathway and education intervention has the potential to improve pediatric asthma management in Alberta. The intervention is anticipated to result in better quality of care for equal or lesser cost.

ClinicalTrial: ClinicalTrials.gov NCT02481037; https://clinicaltrials.gov/ct2/show/NCT02481037 (Archived by WebCite at http://www.webcitation.org/6fPIQ02Ma).

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KEYWORDS

asthma; child; chronic disease management; primary care; clinical pathway; asthma education

Introduction

Background and Overview

Asthma is the most common chronic disease of childhood [1-5]. At least 15% of Canadian children are affected by this disease, and over 50% of these cases are poorly controlled [3-6]. Asthma is among the top 10 reasons for Albertan children to visit an emergency department and the top 5 reasons for urgent admissions to hospital [7,8]. As compared with healthy children, children with asthma miss three times as many school days [9,10] and have substantially reduced quality of life [11,12]. From a societal perspective, inadequate asthma control also presents a considerable economic burden due to the costs associated with hospital and emergency department admissions, medications, and caregiver productivity losses [13].

High-quality evidence exists regarding how to best manage childhood asthma in a primary care setting to optimize control and minimize acute disease exacerbations [14-16]. The best treatment varies depending on the child's asthma disease phenotype, of which there are broadly two types. Although the terms used vary, we refer to these types as "persistent/seasonal disease" and "intermittent/viral triggered" [16-18]. Persistent/seasonal disease has continuous symptoms and is often triggered by allergens, exercise, cold air, and viral illnesses, whereas intermittent/viral-triggered disease is predominately provoked by viral upper respiratory tract infections and resolves completely between infections. For most children with persistent/seasonal disease, inhaled corticosteroids substantially reduce ongoing symptoms, bronchodilator use, asthma exacerbations, and health care utilization [14-17]. For children with intermittent/viral-triggered disease, montelukast (Singulair) has been shown to substantially reduce wheezing exacerbations, asthma symptoms, and health care utilization [19-21].

Although some studies have demonstrated methods to optimize asthma outcomes and minimize costs, this does not guarantee that patients actually receive the best care. It is estimated that across the spectrum of health care, 30-40% of patients do not receive treatments of proven effectiveness [22,23]. Studies from the United States have shown that primary care practitioners do not use preventive medications in up to 45% of children with asthma [24,25], and unpublished data (2008-2011) reported by *Alberta Health Services (AHS)* from Alberta pharmacies show that about 50% of children with asthma who are treated with rescue beta-agonists do not also receive preventative medications [26].

Clinical pathways are a common strategy for changing professional behavior and improving the quality of care delivery, reducing practice variation, and reducing costs of health care [27-30]. Although relatively few published studies have rigorously evaluated the impact of clinical pathways on clinical outcomes and costs [29], it is widely recognized that the rigor involved in pathway development and assessment of evidence obtained is important to improving outcomes. Accordingly, many health organizations such as AHS are using clinical pathways with this intent. In support of this approach, a clinical pathway focused on the emergency management of childhood asthma has been shown to increase the use of evidenced-based therapies [29,30]. However, creation of clinical pathways (or guidelines) is not enough; active implementation strategies are needed to achieve optimal improvements in outcomes [31-34]. Two strategies that are among the most consistently successful for changing practitioner behavior and improving patient outcomes are interactive educational sessions and reminder systems [31-34].

Even if children are prescribed appropriate medications, parents frequently do not receive adequate information and education to understand the rationale and importance of using preventive medications, which often results in poor adherence [35-37]. *Adherence* refers to a patient successfully taking medication. *Primary adherence* refers to the obtaining of medication prescribed, and *secondary adherence* refers to proper completion of the regime as intended by the prescriber. A World Health Organization [38] review indicated 30% adherence to medications by children and adolescents with asthma, which is among the lowest reported for any chronic disease. There is good evidence, however, that asthma education programs for children with severe exacerbations requiring emergency department and in-hospital care sufficiently improve adherence to reduce subsequent hospitalizations, emergency department visits, and urgent disease-related physician visits by 20-30% [39].

We propose to improve the prescription and use of evidenced-based preventative therapies for children with asthma, with the goal to significantly improve their disease control and quality of life, and reduce unnecessary emergency department visits and hospitalizations by 20% each. We will achieve this by (1) installing a primary care clinical pathway for managing childhood asthma into clinicians' electronic medical record

(EMR) to facilitate the use of best evidence by practitioners, and (2) training chronic disease management (CDM) health professionals to provide targeted and timely asthma education to parents and children with asthma. We will test this pathway and education project in a representative sample of 22 Alberta primary care practices, using a pragmatic cluster-controlled trial methodology. Specifically, we will determine if our proposed innovation, as compared with usual care,...

- ...increases *prescription* of evidenced-based preventative therapies;
- ...increases *dispensing* (primary adherence) of evidenced-based preventative therapies;
- ...improves children's *asthma control*;
- ...improves *quality of life* for children and families; and
- ...decreases *health care utilization* (emergent primary care visits, emergency department visits, and hospitalizations) for asthma, and decreases overall societal (health system and family) costs.

Foundational Activities

In Spring 2011, the *AHS* Respiratory Network—now formally known as the "Respiratory Health Strategic Clinical Network (RHSCN)"—adopted the childhood asthma pathway initiative, in collaboration with the *AHS* child health community, as one of its 3 top priorities and provided human and financial resources to move the pathway initiative forward. The initial project of the *RHSCN* and our project team was to develop, implement, and evaluate emergency department and in-patient pathways, for which we received the *Canadian Institutes of Health Research Partnerships for Health System Improvement* funding. Concurrently, with the start of the emergency department/in-patient pilot project and the ongoing implementation of the emergency department and in-patient pathways across the province, the *RHSCN* started developing the primary care pathway.

In Fall 2013, our team developed a collaboration with the *Health Quality Council of Alberta (HQCA)* who have agreed to extract, link, and analyze health administrative data (which they routinely receive and utilize) and other data provided to them from 2 Albertan community-based primary care research networks (Northern and Southern Alberta Primary Care Research Networks—*NAPCReN* [40] and *SAPCReN*, respectively 41). These networks are regional networks contributing to the *Canadian Primary Care Sentinel Surveillance Network* (*CPCSSN*) [42]. *CPCSSN* extracts clinical data quarterly from participating practitioners' (*sentinels*) EMRs, cleans them, removes identifiers, and makes these data available for surveillance, epidemiology, and health services research. With the principal investigator (AC), *CPCSSN* is currently developing and validating a case definition and case finding algorithm for childhood asthma, which will be added in due course to its routine data extraction and processing. The networks contributing to *CPCSSN* in Alberta are currently able to process data from both the *Wolf* and *Med Access* EMR systems. These networks have agreed to assist with recruiting practices to participate in this project.

The project team has engaged extensively with frontline primary care practitioners, primary care leaders, and pediatric respirologists and allergists in developing this primary care pathway project over the last 3 years; each of these groups has heavily invested and is committed to the successful completion of our project. We have engaged frontline practitioners in the following ways: (1) being part of an interdisciplinary pathway development committee including primary care and subspecialty care practitioners with rural, regional, and urban representation; (2) conducting a small focus group (consultation) session about the content and format of the pathway with primary care practitioners at Alberta primary care conferences; (3) beta-testing of the EMR-based pathway in 4 primary care practices; and (4) surveying Alberta primary care practitioners for feedback about the pathway, its design, and practicality. The capacity to manage their asthma patients according to the best practice will be enhanced by the integration of the pathway into the EMRs and by the education given to the CDM health professionals. Participating clinics will identify an individual available for the practice who will take on the role of a CDM professional. The CDM professionals will receive a 3-hour training session/updating on asthma education with continued support from the trainer for the duration of the intervention. They will be provided with access to resources that will be available thereafter to ensure sustainability.

Currently, an asthma case definition is being developed, which is an algorithm that screens the *CPCSSN* data extracted from EMRs to accurately identify patients with asthma. Once developed, the case definition will be validated by comparing it with a gold standard established by 2 experienced physicians using 1000 pediatric *CPCSSN* records.

Network leadership will encourage their members' participation in this initiative by introducing the study team to the medical and administrative leads of all their participating practices. Alongside *SAPCReN* and *NAPCReN* research staff, study team members will travel to eligible practice sites to inform them about the details of the project, answer any questions they might have, and then ask them to participate in this project.

Key Stakeholders

In this project, the following 4 organizations will each play an essential role: AHS (*RHSCN*), *HQCA*, and *NAPCReN/SAPCReN*. *RHSCN* and *HQCA* are committing substantial in-kind funding toward this project over the next 3 years. In addition, 3 other strategic clinical networks, namely, (1) *Maternal, Newborn, Child and Youth*, (2) *Emergency*, and (3) *Primary Care Chronic Disease Management*, share substantial common ground. A total of 3 primary care organizations, the 2 *AHS Primary and Community Care* portfolios and the *Provincial Primary Care Network*, will be "end users," throughout the project, and will also provide critical advice as we plan our "implementation scale and spread" rollout. Participating primary care practices are also end users, providing valuable input into feasibility and sustainability. Finally, the patients of Alberta will be the final stakeholders, as we anticipate their health will be affected favorably.

Methods

Project Objectives

We propose a 3-year mixed-methods health services research project using a randomized cluster-controlled trial design (with primary care practices as the cluster; trial registration number NCT02481037) to achieve the following objectives:

- To use the Theoretical Domains Framework (TDF) to refine our implementation strategy;
- To implement the intervention (a childhood asthma primary care clinical pathway in EMRs and identifying and training CDM health professionals to provide asthma education) in the 11 practices in the intervention arm;
- To evaluate our project intervention using interviews, health administrative and EMR databases, and child and parent surveys of quality of life, the child's asthma control, and the socioeconomic burden of the child's disease on their family; and
- To develop a detailed "scale and spread" strategy if our intervention is shown to be successful (ie, yields either cost savings or improved child and parent quality of life) for all Alberta primary care practices who use a supported EMR.

Intervention Development and Implementation Strategy

The development of our implementation strategy has followed the *Knowledge-to-Action Model* of Graham and colleagues [43]. This cyclic process starts with the identification of the problem—a knowledge-practice gap in the management of childhood asthma in a primary care setting—followed by synthesis of the evidence into a primary care pathway [18], assessment of potential barriers to its use, tailoring of known-effective implementation strategies to local circumstances, evaluation to determine whether the problem has improved and feedback of evaluation results to all end users (decision makers and participating practitioners).

The primary care pathway developed over 2 years by a subcommittee of the Asthma Working Group, RHSCN, was geographically representative of Alberta and included primary care practitioners (n=4), asthma educators (n=5), pharmacists (n=1), allergists and respiratory physicians (n=4), and asthma policy decision makers (n=2). This committee critically reviewed existing global and national guidelines [14,15] and recent randomized trials and systematic reviews to formulate the pathway. Drafts of the pathway were critically reviewed two times by primary care practitioners for ease of use and relevance to their practice, and revised each time.

The following sections provide a summary of the core components of the intervention to be provided to the intervention arm practices of the study:

Pathway Implementation Strategies

Based on previous extensive experience in implementing clinical pathways [44-46] and best evidence available [32,33,47-49], two change strategies were selected to implement the proposed project: (1) an interactive online learning module to teach primary care practitioners about the evidence for treatment strategies in the clinical pathway, and (2) user-written software installed within practitioners' EMR that will serve as a reminder system to facilitate improved patient documentation and management. We will also use the results of a study being conducted using the TDF to identify necessary modifications to our strategy for primary care practices randomized to the intervention arm [50].

Development of EMR Embedded Pathway

The EMR prototype (developed in *Med Access* and currently being developed in *Wolf*) provides practitioners with dropdown menus to document key findings from medical history review and physical examination, classify patient's disease phenotype to generate therapeutic recommendations, assess asthma control, generate a risk assessment, and print out asthma action plans and patient prescriptions.

Development of a Web-Based Interactive Learning Module

To ensure primary care practitioners understand the underlying rationale and evidence behind the development of the primary care pathway, practitioners randomized to the intervention arm will have easy access to a Web-based interactive learning module, which is designed to easily fit into a busy practitioner's schedule. An equivalent module was developed by the research team and successfully used in a trial of an educational intervention in emergency room and in-patient management in Alberta [51]. Physicians and nurses will access the site with a password and will indicate completed sections before the intervention commences. A well-designed Web-based professional learning module has been shown in a previous trial to be as effective at enhancing learning and changing professional behavior as in-person interactive teaching sessions [52]. Parents will also be given access to patient-oriented Web modules to enhance the nurse-led teaching.

Train-the-Trainer for Patient Education

CDM health professionals working within the practices in the intervention arm will be provided 3 hours of in-person training on providing asthma education to children with asthma and their families. This training will be provided by the clinical coordinator, an experienced Certified Respiratory Educator (HS).

Project Population

Practices will be recruited from those who are currently or are becoming a member of the *NAPCReN* or *SAPCReN*. All children (1-17 years of age at the time of enrollment) who meet the case definition of asthma (and are seen by a participating primary care practitioner within a consenting practice) will be included in the trial's analysis unless they refuse consent [53]. The *CPCSSN*s use a "social contract" consent process, where practice populations are informed generally about research, information is freely available on specific projects, and people are included unless they object [53]. Because the project will collect data outside the *CPCSSN* protocol, parents of children with asthma in the participating practices will be sent a letter by their primary practitioner outlining implied consent and asking them to complete surveys about quality of life, asthma control, and the burden (impact) of asthma on their family.

Sample Size Calculation and Analysis

Our *AHS* partners provided the number of pediatric asthma incidences billed by each practice in the province. We will use this information to select practices that have more than 70 active, eligible asthma patients. In addition, the Primary Care Asthma Working Group of clinicians agreed the minimal clinically important difference to be 20% (see Multimedia Appendix 1 for a detailed sample size calculation).

Patients included in the analysis of the primary and one of the secondary outcomes (proportion of children dispensed a preventive medication) will be categorized as either "successful" or "not successful" based on the logic outlined in Multimedia Appendix 2. Given the clustered nature of patients' "success" within practices, a mixed effects logistic regression model will be performed where clustering is considered. Odds ratios for success between arms will be presented along with 95% CIs. The modeling will include assessment of potential confounders and independent risk factors. To analyze the other secondary outcomes (quality of life and asthma control and emergency department visits/hospitalizations), we will use a linear mixed effects regression model, and include fixed effects for the baseline/follow-up periods, the pathway/control arms, and stratifications/interaction, and random effects to account for clustering in practices.

Randomization of Practices Design Issues and Potential Bias

All practices consenting to participate in this trial will be randomized, stratified for urban (Edmonton/Calgary) versus nonurban and academic (trainees routinely evaluate patients) versus nonacademic practices. A statistician not aware of our project question will allocate practices using a standard randomization software. We have chosen practices as the unit of randomization because most individual practitioners within a shared practice use the same EMR and we wish to prevent contamination. Because we are testing the effectiveness of embedding the pathway into clinicians' EMR and train-the-trainer CDM professional education, consenting practices randomized to usual care will not have access to either intervention, and therefore the risk of significant contamination is small. In addition, the design provides an opportunity to test for contamination by comparing the difference between the pre- and post-periods for the 2 groups (ie, intervention and control).

Practices will be randomized to either:

- Routine care versus intervention
- Embedding a primary care clinical pathway for managing childhood asthma into clinicians' EMR to facilitate practitioners utilizing best evidence, and training these practices' CDM professionals to provide asthma education to children with asthma and their parents.

Quantitative Data

Data will come from 3 sources, namely, health administrative sets, data extracted from participating practices' EMR, and surveys of asthma control, quality of life, and socioeconomic factors. Health administrative datasets will include Alberta Patient Registry, emergency department visits (*National Ambulatory Care Reporting System*), hospitalization abstracts (*Discharge Abstract Database*), practitioners visits (*Alberta physician/NP claims*), and pharmacy dispensing (*Pharmaceutical Information Network*); these datasets are considered to be of good quality with accurate personal identifiers on more than 95% of records. As previously noted, EMR data using a validated case definition will be extracted for all children with asthma meeting the inclusion/exclusion criteria. This will provide individual categorical and continuous patient-level data such as patient demographics, encounter history, medication prescriptions, and comorbid disease. Also as previously noted, the *HQCA* will play a pivotal role in our trial as they will (1) receive the aforementioned datasets from *AHS*, Alberta Health, and *NAPCReN/SAPCReN*; (2) extract and link the necessary data using a separate file from the practitioners, which will contain patients' *CPCSSN* identifier linked to their Alberta Health Care Number; (3) perform the planned analysis; and (4) provide the aggregated results to our research team.

Letters from participating practitioners will be sent two times to families of children with asthma before and 11 months after the study intervention starts. The letter will inform parents about the purpose of the survey (though the broad purpose of the project will not be revealed to maintain masking) and will ask the primary parent or caretaker to go online to review the information sheet and complete a socioeconomic survey and the *Paediatric Asthma Caregiver's Quality of Life Questionnaire (PACQLQ)* for children with asthma, or to complete written copies of the questionnaires and return them by self-addressed stamped envelopes. A previous study [54] has validated the *PACQLQ* for use by parents of children less than 7 years of age. Children 7 years and older will be asked to also complete the *Standardised Paediatric Asthma Quality of Life Questionnaire* and the *Asthma Control Questionnaire* [55]. The socioeconomic survey includes questions such as day care or school days missed, parent work days missed, and asthma medications purchased in the last 30 days, to allow calculation of the economic impact of a child's asthma on their family.

Qualitative Data

We will purposefully select 6 practices randomized to the pathway and education intervention based on size of the practice and heterogeneity of professionals in the practice [56], and evaluate them using multidisciplinary focus groups after the intervention. The interview protocols will be developed following the implementation of the intervention to incorporate key learning from the implementation into the question development. The focus groups will include practitioners and other professionals, for a total of at least six clinicians, and will be recorded by a court reporter [57]. The content of the interview questions will be guided by the Knowledge to Action Model [58] and will target areas such as attributes of the pathway, adopters, and implementation setting. Clinicians will have the opportunity to provide feedback regarding potential challenges with the pathway and offer suggestions for improvements.

To monitor the progress and permit follow-up of ideas that emerge from the focus group, data collection and analysis will follow an iterative and concurrent process. The

post-implementation focus group data will be analyzed in the following three phases: coding, categorizing, and developing themes [59]. First, data for each practice will be coded to facilitate analysis. The code word(s) will reflect the essence of the data leading to ease of recognition as the number of code words increases. Codes will be operationally defined so that they can be consistently applied throughout the data. Second, codes will be placed into broad categories that correspond to the major unit of analysis. As categories emerge, their theoretical properties will be defined. Third, comparisons between categories will be carried out to locate similarities and differences among them. Data analysis will be carried out using NVivo 10.

Economic Analysis

The potential for provincial-wide savings is achievable because of the high-quality evidence (numerous randomized controlled trials, systematic reviews, and international consensus guidelines) demonstrating that appropriately tailored anti-inflammatory medications reduce emergency department visits and hospitalizations and that asthma education training targeted to focus on families with children with more severe disease also yields a 20% reduction in subsequent hospitalizations, a 30% reduction in emergency department visits, and a 30% reduction in nonscheduled urgent physician visits for disease exacerbation [39]. Further validating the ability of preventative therapy for asthma to reduce health care utilizations and costs, health services studies in children and adults in both British Columbia and Sweden utilizing administrative data have shown that higher utilization of inhaled corticosteroids is correlated with lower rates of hospitalization and lower overall health care costs [60,61].

The primary analysis will be from the health care system perspective. Total health system costs per child with asthma in the 2 trial arms before and after the intervention will be determined. Health system costs will be disaggregated into the following three parts: (1) those stemming from pathway implementation and train-the-trainer education sessions, 2) those from asthma education provided by CDM professionals, and (3) those from hospitalizations and emergency department and practitioner visits for asthma. Overall health system costs will be considered decreased by the project intervention if the cost decrease from reduced emergent physician visits, emergency department visits, and hospitalization outweighs the cost increase from pathway implementation and asthma education. The impact on costs will be considered alongside the impact on outcomes including quality of life. A secondary analysis will also include consideration of family costs that consist of parent loss of work, travel, or other out-of-pocket costs due the child's disease.

Costs stemming from health care utilization (drugs, hospitalizations, emergency department, and practitioner visits) will be derived from health administrative data. Actual costs are available from *AHS* for in-patient stays at Calgary and Edmonton Zone hospitals, and costs for other Alberta hospitals can be approximated using Institute of Health Economics (IHE) provincial estimates [62]. Emergency department visit costs will be determined using the IHE standard provincial estimates [62]. Surveys of staff and parents of children with asthma from each of the participating practices will provide cost estimates by collecting the following data: time spent on train-the-trainer sessions, staff time to educate families, and family quality of life measures, as well as out-of-pocket expenditures on their child's disease.

Ethics Approval

The protocol obtained approval from the Health Research Ethics Board at the University of Alberta (Edmonton, AB) on August 7, 2015, and the Conjoint Health Research Ethics Board at the University of Calgary (Calgary, AB) on September 23, 2015.

Results

Practice recruitment began September 2015, and all study-related activities are expected to conclude March 2018 (see Multimedia Appendix 3 for a timeline of major objectives) with the following outcomes:

Primary Outcome

The proportion of symptomatic children with asthma in the baseline and follow-up periods (separate calculations) who are appropriately treated with a preventer will be the primary outcome. The denominator will be the number of children who meet the case definition of asthma *and* receive at least one prescription for an inhaled short-acting beta-agonist (eg, salbutamol) during the applicable period. The numerator will be, of these children, the number of those who receive a prescription for inhaled corticosteroid, montelukast, a combination of inhaled long-acting beta-agonist and corticosteroid (ie, agonist + corticosteroid), or some combination of these 3 drugs in the same period.

Secondary Outcomes

Dispensed Preventative Therapies (Primary Adherence)

The proportion of applicable children in baseline and follow-up periods who are appropriately *dispensed* a preventer will highlight if there is a significant gap between prescriptions given and filled. The denominator will be the same as for the primary outcome but the numerator will be the number of these children who are *dispensed* 1 or more preventer medications from newly available PIN data.

Emergency Department Visits and Hospitalizations for Asthma

The number of asthma emergency department visits or hospitalizations for asthma (ICD10 J45 or J46) per child who meets the case definition of asthma during each period will be a measure of health care use.

Electronic Medical Record Data

The *CPCSSN* and research team will develop and validate a case definition and case finding algorithm for identifying children with asthma in the *CPCSSN* practices. The *CPCSSN* EMR data will provide, for all eligible children, individual categorical and continuous patient-level data such as patient demographics, ICD9 codes, and medication prescriptions. For children evaluated in practices randomized to the pathway group,

data will also include asthma phenotype and provision of asthma action plans.

Discussion

The management of pediatric asthma in Alberta is variable in quality with only half of the affected children having control of their asthma. This has adverse effects on morbidity and costs of health care. We aim to improve the quality of care in primary care by a two-pronged approach: the use of a management pathway in the practitioner's EMR and the education of parents in the importance of preventive therapies.

At the end of the 3-year project, the rigorous pragmatic cluster-controlled design will provide a thorough and comprehensive understanding of whether this project improves patient care, how effective the interventions are, and whether the interventions add value or are cost saving. If the results are positive, we will collaborate with the *AHS RHSCN*, the *Provincial Primary and Community Care* portfolios, and the *Provincial Primary Care Network* to deliver this pathway and educational initiative to all Alberta primary care practices and their patients.

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Conflicts of Interest

None declared.

Multimedia Appendix 1

Sample size sensitivity analysis.

[PDF File (Adobe PDF File), 38KB - resprot_v5i1e37_app1.pdf]

Multimedia Appendix 2

Logic model for determining â€˜overall successâ€™ (appropriate treatment of symptomatic asthmatic child with preventer medication) for primary outcome.

[PDF File (Adobe PDF File), 27KB - resprot_v5i1e37_app2.pdf]

Multimedia Appendix 3

Timeline of major objectives and associated milestones.

[PDF File (Adobe PDF File), 148KB - resprot_v5i1e37_app3.pdf]

Multimedia Appendix 4

Peer-review from funding agency.

[PDF File (Adobe PDF File), 15KB - resprot_v5i1e37_app4.pdf]

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Abbreviations

AHS: Alberta Health Services
CDM: chronic disease management
CPCSSN: Canadian Primary Care Sentinel Surveillance Network
EMR: electronic medical record
HQCA: Health Quality Council of Alberta
IHE: Institute of Health Economics
NAPCReN: Northern Alberta Primary Care Research Networks
PACQLQ: Paediatric Asthma Caregiver's Quality of Life Questionnaire
RHSCN: Respiratory Health Strategic Clinical Network
SAPCReN: Southern Alberta Primary Care Research Networks
TDF: Theoretical Domains Framework

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Original Paper

Increasing Antiretroviral Adherence for HIV-Positive African Americans (Project Rise): A Treatment Education Intervention Protocol

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Abstract

Background: HIV-positive African Americans have been shown to have lower adherence to antiretroviral therapy (ART) than those of other races/ethnicities, yet adherence interventions have rarely been tailored to the needs of this population.

Objective: We developed and will evaluate a treatment education adherence intervention (called Rise) that was culturally adapted to address the needs of African Americans living with HIV.

Methods: This randomized controlled trial will examine the effects of the Rise intervention on ART adherence and HIV viral load. African Americans on ART who report adherence problems will be recruited from the community and randomly assigned to receive the intervention or usual care for 6 months. The intervention consists of 6-10 individual counseling sessions, with more sessions provided to those who demonstrate lower adherence. Primary outcomes include adherence as monitored continuously with Medication Event Monitoring Systems (MEMS) caps, and viral load data received from the participant's medical provider. Survey assessments will be administered at baseline and month 6.

Results: The trial is ongoing.

Conclusions: If effective, the Rise intervention will provide community-based organizations with an intervention tailored to address the needs of African Americans for promoting optimal ART adherence and HIV clinical outcomes.

Trial Registration: Clinicaltrials.gov NCT01350544; https://clinicaltrials.gov/ct2/show/NCT01350544 (Archived by WebCite at http://www.webcitation.org/6fjqqnmn0).

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KEYWORDS

antiretroviral treatment; adherence; patient compliance; HIV; African Americans; treatment education; intervention

Introduction

Compared to Whites living with HIV, African Americans with HIV have lower engagement and retention in care [1-3], are less likely to receive antiretroviral treatment (ART) [4,5], and are less likely to adhere to treatment long enough for it to be effective [4-9]—all of which may contribute to disparities in survival [10-13].

Research has identified culturally specific determinants of ART adherence among African Americans, including stigma, medical mistrust, and HIV-related misconceptions (eg, that ART is poison) [7,8,14,15]. Research also suggests that social conditions such as poverty, health care factors (eg, provider behaviors contributing to mistrust), and psychosocial issues such as mental health need to be addressed in interventions focused on improving HIV-positive African Americans' health behaviors and ultimately their health outcomes [16-19]. However, interventions to improve adherence have rarely been culturally tailored to the needs of African Americans, which may partially explain the lack of robust effects observed in reviews of ART adherence intervention trials that contain large numbers of African American participants [20-22].

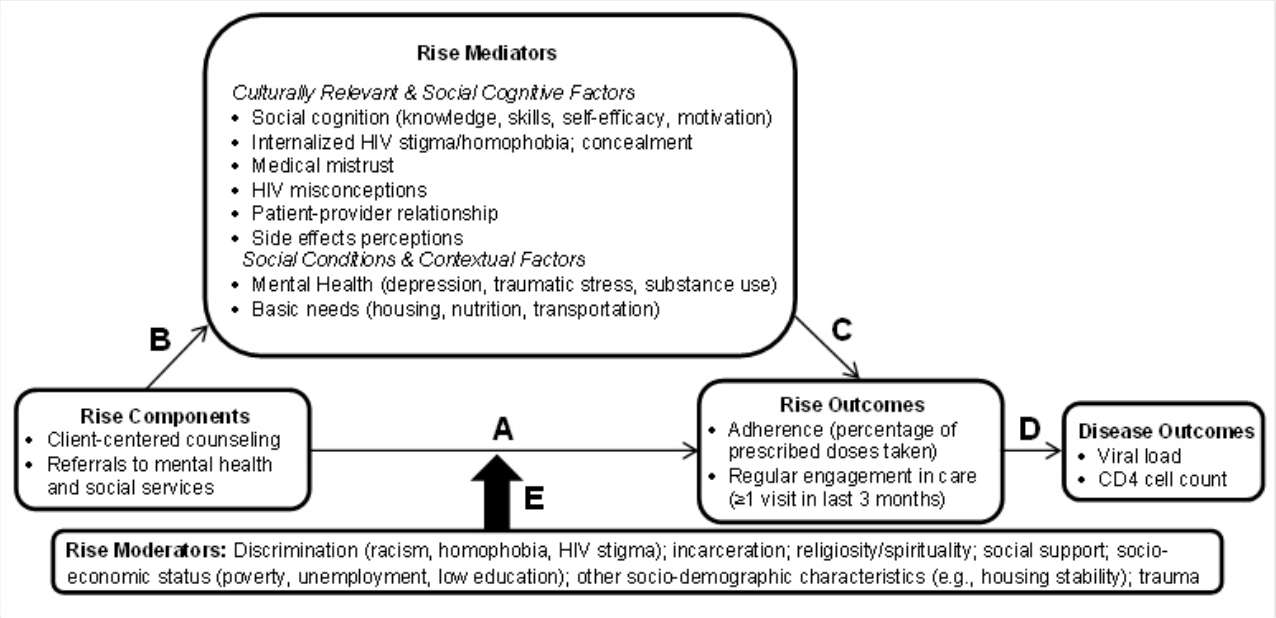
Treatment education programs have been used by AIDS service organizations (ASOs) across the United States to facilitate adherence to HIV care through client-centered one-on-one counseling. Treatment education counselors possess specialized HIV treatment knowledge, target structural issues in health care and social conditions in clients' lives, counsel clients to overcome adherence barriers, recommend changes in treatment and/or providers (if needed), and refer clients to mental health, substance abuse, and social services (eg, for housing). Treatment education is particularly appropriate for patients who may be mistrustful of providers, including African Americans, because it can be conducted outside of the medical system, and teaches patients self-advocacy in health care.

To our knowledge, the efficacy of treatment education has not been evaluated in a randomized controlled trial (RCT). In a prior process evaluation of the longstanding treatment education program at AIDS Project Los Angeles (APLA), clients in treatment education showed higher adherence and engagement in care, greater perceived ART efficacy, and fewer unmet social service needs, compared to clients not receiving treatment education [23]. Furthermore, treatment education was viewed by medical providers, treatment education counselors, and patients as increasing understanding about treatment, support adherence, and improving patient-provider relationships [24].

We developed the structured, culturally tailored treatment education intervention *Rise*, named by the community-academic team after the Maya Angelou poem [25] "Still I Rise," which emphasizes resilience in Black communities. We are evaluating Rise's effects on adherence among African Americans in an RCT, the protocol for which is described in this paper. Rise is built around the core components of the treatment education program being implemented by APLA, which include a needs assessment, individual counseling, and referrals as required. These components are proposed to synergistically influence sociocultural factors that predict adherence (see Figure 1). Client-centered counseling builds treatment knowledge and adherence skills, self-efficacy, and motivation (key adherence predictors) [26-28]. Given robust relationships of homelessness, depression, traumatic stress, and substance use with adherence [29-30], referrals to mental health/social services can improve adherence by decreasing unmet needs. Rise also addresses culturally relevant coping strategies (eg, medical mistrust) that arise in response to factors such as discrimination and stigma, which can undermine health care access and use in Black communities [31,32].

This paper describes in detail the methodological protocol, intervention content, and structure of Rise. If effective in improving ART adherence and virologic suppression, Rise will provide the field with one of the first ART adherence interventions tailored specifically to address the needs of African Americans.

Figure 1. Conceptual model of Rise, a culturally tailored treatment education program.



Methods

Study Design

The study is an RCT to compare Rise, a culturally tailored and structured adherence counseling intervention, with usual care (received from participants' primary care providers, with no added intervention), among African Americans living with HIV. To determine effects on ART adherence, participants complete audio computer-assisted self-interviews (ACASI) at baseline and 3- and 6-months postbaseline to assess self-reported adherence, and interviewers check in with participants at 1-, 2-, 4-, and 5-months postbaseline to download electronically monitored adherence data, collected via the Medication Events Monitoring System (MEMS) described below, and to update contact information. Participants are paid $30 at baseline and 3- and 6-month follow-ups, and $10 for each check-in (1, 2, 4, & 5 months postbaseline). If all assessments are completed, participants receive a $20 bonus. Participants also receive a second $20 bonus for updating primary contact information during the study (eg, address, phone, email). Participants receive a snack and $10 per intervention session for transportation costs. The Human Subjects Protection Committee of the RAND Corporation and the Institutional Review Board of California State University, Dominguez Hills approved the study protocol, and a Certificate of Confidentiality was obtained from the National Institutes of Health. The trial is registered with the National Institutes of Health sponsored clinical trials registry and assigned the identifier NCT01350544 (date released: 5/26/2011).

Study Setting

The study is being conducted at AIDS Project Los Angeles (APLA), the largest ASO in Los Angeles County. APLA provides direct, bilingual services (eg, treatment education, case management, substance use and mental health treatment, housing and food assistance) to ~10,000 men, women, and children with HIV in Los Angeles county annually. The majority of APLA clients are ethnic or racial minorities, with about one-quarter being African American, and nearly 90% male, which is consistent with the racial/ethnic and gender distribution of people living with HIV in Los Angeles.

The study team represents a partnership among researchers, APLA program staff, and HIV community stakeholders including a community advisory board (CAB) composed of clients living with HIV (most of whom are African American), APLA staff, and HIV service agency and staff. This has played a pivotal role in the study design and suggested directions for the intervention. Throughout the ongoing RCT, CAB members have been integral in reviewing project methodology and materials, evaluating study progress, and monitoring program implementation; when the results are available, they will also help with interpreting study data. Eliciting the feedback of the CAB members is facilitated by in-person CAB meetings that are convened at key junctures of the study, about two to three times during the course of each year of the study. CAB members are given a small honorarium for their time ($50 per meeting).

Participants

Eligibility criteria include the following: (1) age ≥18 years; (2) self-identification as African American or Black (if mixed race, primarily identify as African American); (3) on ART; (4) reported any adherence problems (ie, missed ≥1 ART dose in the past month, less than 100% adherence in the past month, sometimes stopped ART if they felt worse, and/or any missed doses during the past weekend); (5) not currently participating in another treatment adherence intervention; and (6) willing to use an electronic monitoring cap to monitor adherence to one of their antiretrovirals. Individuals who have received treatment education services (from APLA or elsewhere) in the last 6 months are excluded. Participants are recruited through APLA and other community-based organizations (eg, SPECTRUM, OASIS) via public presentations of the study and flyers, referrals from providers, and radio and print advertisements.

Treatment Advocacy Intervention (Rise)

The 24-week intervention consists of 4 individual, weekly, 60-minute adherence counseling sessions and 1 group HIV education session during the first month, which comprises the core adherence training phase. This is followed by a maintenance phase, in which participants receive booster sessions at weeks 12 and 20, plus up to 4 additional sessions depending on adherence performance (ie, sessions added if <90% of prescribed doses taken in past month). In total, participants receive between 6 and 10 individual sessions. Table 1 describes the intervention components, goals, and activities of each session, and the mediators of intervention effects that are addressed by these activities. Table 2 provides an outline of the exercise content within each session.

Table 1. Rise session components, goals, activities, and proposed mediators addressed.

Rise Component/Goal	Individual Session	Counselor Activity	Mediator Addressed
Client-centered Counseling: Improve motivation for adherence and care engagement through use of MI[a] style and provision of strategies to reduce barriers	1	After introducing the program, assess client's knowledge and provide education on HIV, ART[b], adherence, and health disparities; discuss culturally relevant stressors (eg, discrimination) that can affect health and health behaviors	Improved adherence cognitions (knowledge, skills, self-efficacy, motivation); lower HIV misconceptions & medical mistrust
	All	Review electronic adherence data, offer encouragement and reinforce good adherence; in all sessions after Session 1, assess changes in engagement in care and adherence	Improved adherence cognitions
	All	Discuss attitudes and beliefs, including culturally relevant health beliefs; encourage positive attitudes toward treatment	Lower HIV misconceptions & mistrust
	All	Assess support availability, and level of stigma and awareness of HIV status in the network; problem solve ways to increase support	Greater HIV disclosure
	2-10	Use steps for problem solving to identify barriers to engagement in care and adherence, and plan solutions	Improved adherence cognitions
	2-10	Discuss integration of medication and appointments into daily routine	Improved adherence cognitions
	2-10	Discuss strategies to reduce/manage medication side effects	Reduction in perceived side effects
	All	Develop (in Session 1) and review (in other sessions) Individual Service Plan (ISP) of short- and long-term goals; proposed timeline and outcomes; and client and medical provider tasks	Improved adherence cognitions
Mental Health/Social Services Referrals: Link to services to improve mental health/reduce unmet needs, to support adherence and care engagement	All	Conduct needs assessment and address problem areas, including social and structural issues (eg, substance abuse/mental health, housing/nutrition) that influence adherence; provide needed referrals	Fewer unmet basic subsistence needs; fewer/less severe mental health symptoms (depression, traumatic stress); lower substance use
Facilitated Group Education: Provide basic education; use group dynamic to increase treatment motivation and in turn, adherence and care engagement	Between 1 & 4	Provide basic information on prevention, transmission, progression, treatment, and adherence; encourage group discussion and address any HIV-related misconceptions	Improved adherence cognitions, lower HIV misconceptions and medical mistrust
	Between 1 & 4	Present information on HIV and adherence disparities, and racism, homophobia, and HIV stigma as contributors to disparities; encourage sharing of health care experiences	Lower HIV misconceptions, medical mistrust, and internalized stigma
	Between 1 & 4	Promote collective responsibility to motivate adherence (eg, stay healthy to keep Black communities strong)	Improved adherence cognitions

[a] motivational interviewing

[b] antiretroviral therapy

Table 2. Outline of intervention session content.

Session 1 (Week 00)	1. Check in with the client to begin to build rapport. 2. Introduce the intervention and its goals. 3. Review client's history of HIV diagnosis and ART, and relationship with provider. 4. Provide education about goals of ART and importance of adherence. 5. Assess client's attitudes towards treatment and adherence. 6. Conduct needs assessment. 7. Develop client's Individual Service Plan (ISP).
Session 2 (Week 2)	1. Check in with client to build rapport. 2. Review progress with ISP since last session and follow up on referrals made. 3. Review adherence using the MEMS print out. 4. Identify barriers to adherence. 5. Introduce problem solving steps. 6. Apply problem solving steps to one barrier. 7. Tailor regimen and adherence to daily routine cues. 8. End with a statement of affirmation.
Session 3 (Week 4)	1. Check in with client to build rapport. 2. Review progress with ISP since last session and follow up on referrals made. 3. Review adherence since last session using the MEMS print out. 4. Identify barriers to adherence. 5. Apply problem solving steps to one barrier. 6. Enhance social support for adherence. 7. Revisit attitudes towards treatment and adherence. 8. Discuss plans to interact with client's HIV provider. 9. End with a statement of affirmation.
Maintenance Module A/B (Week 12/20)	1. Check in with client to build rapport. 2. Review progress with ISP since last session and follow up on referrals made. 3. Review adherence over past month using the MEMS print out. 4. Identify barriers to adherence. 5. Apply problem solving steps to one barrier. 6. Enhance social support for adherence. 7. Revisit attitudes toward treatment and adherence. 8. Review status of doctor-patient relationship. 9. End with a statement of affirmation.
Last Session Add-Ons	1. Instill confidence and self-efficacy for long-term adherence. 2. Set goals related to adherence over the next few months.

Core Adherence Training Phase

Session 1 (Week 1)

In this first session, the counselor provides information about the importance of adherence and consequences of missed doses, provides education about viral load and drug resistance, and explains the connection between dosing schedules and viral suppression. A motivational interviewing (MI) style [33] is used to help clients develop or strengthen positive attitudes toward treatment and adherence. We do not attempt to adhere strictly to the use of MI, but the counselor is trained to ask open questions, to use reflective listening, and to motivate change by highlighting discrepancies between behavior or thoughts and stated health goals, as well as respecting client autonomy. Following a needs assessment, the counselor provides referrals for any unmet basic needs (housing, food, transportation) and mental health issues (depression, traumatic stress, substance use).

To address culturally relevant factors, the counselor acknowledges health care discrimination and health disparities, and shares reasons for medical mistrust and HIV misconceptions in Black communities. The counselor encourages the client to share any experiences and perceptions they may have in this regard. To enhance instrumental (eg, finding transportation to clinic) and emotional (eg, encouraging/reinforcing adherence) support, the counselor assesses the availability of social supports to the client (eg, whether individuals in clients' social networks are aware of their HIV status). The counselor explores with the client whether HIV disclosure is an option for obtaining social support, but in the context of reviewing both possible benefits as well as risks to disclosure. If the client reports HIV-related misconceptions, the counselor supplies accurate information to dispel the misconceptions.

To diminish mistrust, the counselor works to establish a collaborative relationship in which the client can experience the counselor as an equal, identify commonalities, and be regarded as a whole person [34]. This may involve using nontechnical or vernacular language when appropriate, and allowing time for clients to discuss issues not directly related to the intervention aims (to demonstrate investment in the client as a person). The counselor maintains a nonjudgmental attitude, to help clients feel safe revealing gaps in their HIV treatment knowledge and reasons for missed doses. At times, the counselor may also reflect on his or her own experiences. For example, when addressing conspiracy theories about HIV, the counselor may share if he or she originally held similar beliefs, and how these views changed through learning more about the relevant research. Sharing these more personal experiences helps the client view the counselor as a "genuine" person who is speaking based on his or her authentic values and feelings—rather than merely performing a professional role. This is also intended to reduce mistrust by modeling critical thinking and receptivity to scientifically based information.

Session 1 closes with the development (in partnership with the client) of an Individual Service Plan (ISP) of short- and long-term goals (eg, following up on referrals, engaging a friend to seek support for adherence), which is revisited and updated in each successive session.

Sessions 2-4 (Weeks 2-4)

These sessions are similar in content to Session 1, but with greater targeting of barriers to medication adherence and retention in care. Together with the client, the counselor reviews adherence (using the printout from the MEMS software, which provides adherence summary statistics and graphic depictions of patterns of doses taken and missed) and attendance at scheduled doctor's visits since the last session. The client identifies barriers that may contribute to missed appointments and nonadherence, such as lack of accurate treatment information (or belief in misconceptions), low motivation, presence of or concern about treatment side effects, mental health problems or substance use, internalized stigma, and medical mistrust and/or negative experiences with health care (eg, discrimination). Stages of problem solving are reviewed: defining the problem, deciding on a goal, generating possible solutions, selecting a potential solution, planning the solution's implementation, and evaluating the solution's effectiveness (at the subsequent session).

The counselor identifies contextual cues that influence pill-taking (ie, what occurred immediately before and after a missed dose/appointment), to derive strategies for managing and controlling cues. Clients describe daily routines and together with the counselor determine optimal ways to integrate medication into these routines. Doses are connected with routine daily activities that can serve as reminder triggers for taking medications. Strategies for coping with and reducing side effects are discussed. The counselor assesses whether clients followed up on referrals, offers new referrals as needed, and evaluates progress toward ISP goals. Within these early sessions, the counselor assesses the client's relationship with their HIV care provider in terms of satisfaction with care received and level of support from and trust in their provider. With consent from the client, the counselor contacts the provider via phone or email to inform them that the client is participating in the program and working with the counselor over the next 6 months to support the client's adherence and care retention.

Group HIV Session

Before Session 4, clients are offered an hour-long HIV education group, facilitated by the counselor, along with other study participants who are receiving the intervention. The counselor provides basic HIV and ART information from APLA's standard educational forum, facilitates discussions of how stigma, discrimination, and medical mistrust contribute to health disparities in Black communities, and encourages participants to share experiences with and learn from each other.

Maintenance Adherence Training Phase

Booster Sessions 5 & 6 (Weeks 12, 20)

The goal of the maintenance phase is to help clients sustain optimal adherence. Clients who achieve ≥90% adherence (as measured by the MEMS cap) during the 2 weeks preceding Weeks 12 and 20 receive a session only at those weeks; others receive up to two biweekly added sessions at each time point. For example, if adherence from Weeks 10-12 is ≥90%, the client receives a session at Week 12, and then returns again at Week 20 for the next booster session. If the client's adherence from Weeks 10-12 is <90%, the client receives maintenance sessions at Weeks 12 and 14, and if adherence from Weeks 12-14 is <90%, then s/he receives another session at Week 16. All clients return at Week 20, but only clients with adherence <90% during Weeks 18-20 return at Week 22, and those with adherence <90% during Weeks 20-22 have another session at Week 24. We used 90% adherence as the cutoff for determining good adherence based on research suggesting that this level of adherence was associated with optimal treatment response and virologic suppression [35]. This dose regulation (of 0-4 extra sessions) is intended to promote efficient use of limited community resources by varying intensity depending on client need. Maintenance sessions are similar to the initial core sessions, with continued emphasis on identification and resolution of barriers, use of motivational enhancement techniques to improve attitudes and motivation related to adherence, side effect management, and monitoring of adherence-related social support and self-efficacy, and medical mistrust.

Intervention Counselor Training, Fidelity Monitoring, and Supervision

Counselor training and ongoing supervision is used to ensure consistency in intervention implementation and monitor fidelity to the intervention protocol. The counselor training includes instruction on basic HIV disease information, the importance of protecting confidentiality and complying with Health Insurance Portability and Accountability Act (HIPAA) regulations, crisis intervention, referral resources, cultural and social issues that can influence treatment adherence among African Americans, and mental health and substance abuse assessment. The counselor is trained to establish rapport and effective working relationships with clients and medical providers, and locate community resources, referrals, and

linkages. The counselor is provided with and encouraged to further develop a comprehensive referral list to HIV-related and auxiliary services (eg, housing, mental health). All counseling sessions are audio-recorded, with consent from the client. The supervisor listens to all recorded sessions of the first 2 clients receiving the intervention, and then all recorded sessions of every 5th participant. This serves as the basis for provision of feedback to the counselor during biweekly supervision. Supervision also provides a time for the counselor to discuss challenging cases and review treatment plans for newly enrolled participants.

Measures

Below are descriptions of the primary outcomes to be assessed (ART adherence, HIV care retention, and HIV clinical outcomes). The survey assessment includes measures of potential mediators of intervention effects (eg, attitudes and beliefs related to HIV, ART and adherence, stigma, discrimination, mental health and substance use), and potential moderators (eg, socio-demographic characteristics); however, these are not described in detail here.

ART Adherence

Adherence to one antiretroviral (with the most complex regimen) is measured electronically and continuously throughout the study using the MEMS. Clients are instructed to remove one dose at the time that they plan to ingest the dose, and to refill the bottle after the last pill is removed. Participants do not always follow these instructions (eg, removing multiple doses at once or "pocketing"); thus, we assess their actual use of the cap via self-report in the past 2 weeks at each primary assessment, and then adjust the number of prescribed does taken during this period using this information. This adjustment method has been validated [35] and shown to strengthen the relationship between adherence and viral load. MEMS software calculates several adherence parameters including the percentage of scheduled doses taken, and yields printouts of adherence data and charts that depict adherence patterns (which are used to facilitate the adherence counseling sessions). We also ask participants to self-report the number of doses missed in the last week and percentage of prescribed medications taken in the last month.

HIV Care Retention

Clients are asked to report the number of appointments scheduled, attended, and missed with their HIV care provider in the past 6 months.

HIV Clinical Outcomes

Clients are asked to provide HIPAA-compliant informed consent for access to medical records data, which we are using to request the last 2 HIV viral load and CD4 count assay results from providers.

Statistical Analysis

To examine the effects of Rise on the primary outcomes, we will use ordinary least squares regression for continuous outcomes (mean adherence; change in viral load and CD4) and logistic regression for dichotomous outcomes (adherence >90%; undetectable viral load). Group assignment (intervention or control) will be entered in the model as a dummy variable, together with the baseline value of the outcome and covariates. For comparability across outcomes, we will choose a single set of covariates across models, from variables hypothesized based on theory and prior research to predict outcomes, or for which we find support for hypothesized relationships in bivariate baseline analyses. We will drop predictors for which there are large correlations to avoid collinearity. We will use an intent-to-treat approach as the primary analysis, grouping participants by the condition to which they were randomized, regardless of level of participation or study completion; a secondary analysis will be performed with study completers only.

Results

The project was funded in 2012 and enrollment was completed in 2015. Data analysis is currently underway and the first results are expected to be submitted for publication in 2016.

Discussion

This study will be one of the first to evaluate an HIV treatment adherence intervention for African Americans that integrates culturally relevant factors with basic components of treatment education, an established program commonly found in AIDS service organizations. Although many HIV adherence intervention trials have included a substantial number of African American participants, few were developed specifically for African Americans and take into account their unique cultural context. Interventions to improve African Americans' adherence have rarely considered cultural and social determinants of health behavior, including medical mistrust, HIV misconceptions, and experiences with discrimination [36].

Although not specific to adherence, meta-analyses indicate that most HIV-prevention interventions for women are less consistently effective for African Americans [37], and interventions addressing culture tend to have larger effects [38]. Further, most adherence interventions are conducted in HIV primary care clinics [39]. For greater success, adherence programs need to take into account African Americans' mistrust of "outsiders" (which limits acceptance of HIV care), openly acknowledge factors such as racism that undermine health care in Black communities [40,41], and address mistrust and stigma as coping strategies that arise in response to racism and oppression. Moreover, given African Americans' high levels of medical mistrust, community-based programs such as Rise may have greater reach and effectiveness than those in clinics.

In sum, Rise integrates current practice with science by examining whether an untested yet sustained community program is effective. Our program also challenges a growing policy movement in HIV clinical care that seeks to engage patients for social services via medical providers (within the confines of the medical system) rather than community settings. Rise's community-based paradigm provides an alternative source of treatment support, which may be critical for African Americans who exhibit high levels of mistrust and low adherence, and for whom no culturally relevant rigorously tested

adherence intervention exists. In this way, Rise is a holistic program that includes client-centered treatment education to intrinsically motivate treatment behavior changes, and referrals to auxiliary services targeting unmet social service needs to ultimately improve health outcomes.

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Authors' Contributions

GW, LB, MM, BM, and BR contributed to the design of the study and its protocol, the development of the intervention, and the writing of this paper. KM contributed to the development of the intervention.

Conflicts of Interest

None declared.

Multimedia Appendix 1

Summary statement from NIH review.

[PDF File (Adobe PDF File), 88KB - resprot_v5i1e45_app1.PDF]

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Abbreviations

ACASI: audio computer-assisted self-interviews
AIDS: acquired immune deficiency syndrome
APLA: AIDS Project Los Angeles
ART: antiretroviral therapy
ASOs: AIDS service organizations
CAB: community advisory board
HIPAA: Health Insurance Portability and Accountability Act
HIV: human immunodeficiency virus
ISP: Individual Service Plan
MEMS: Medication Event Monitoring Systems
MI: motivational interviewing
RCT: randomized controlled trial

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Protocol

Online Support Program for Parents of Children With a Chronic Kidney Disease Using Intervention Mapping: A Development and Evaluation Protocol

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Abstract

Background: The care for children with a chronic kidney disease (CKD) is complex. Parents of these children may experience high levels of stress in managing their child's disease, potentially leading to negative effects on their child's health outcomes. Although the experienced problems are well known, adequate (online) support for these parents is lacking.

Objective: The objective of the study is to describe the systematic development of an online support program for parents of children with CKD, and how this program will be evaluated.

Methods: Intervention Mapping (IM) was used for the development of the program. After conducting a needs assessment, defining program objectives, searching for theories, and selecting practical applications, the online program e-Powered Parents was developed. e-Powered Parents consist of three parts: (1) an informative part with information about CKD and treatments, (2) an interactive part where parents can communicate with other parents and health care professionals by chat, private messages, and a forum, and (3) a training platform consisting of four modules: Managing stress, Setting limits, Communication, and Coping with emotions. In a feasibility study, the potential effectiveness and effect size of e-Powered Parents will be evaluated using an explorative randomized controlled trial with parents of 120 families. The outcomes will be the child's quality of life, parental stress and fatigue, self-efficacy in the communication with health care professionals, and family management. A process evaluation will provide insight in parents' experiences, including their experienced level of support.

Results: Study results are expected to be published in the summer of 2016.

Conclusions: Although the development of e-Powered Parents using IM was time-consuming, IM has been a useful protocol. IM provided us with a systematic framework for structuring the development process. The participatory planning group was valuable as well; knowledge, experiences, and visions were shared, ensuring us that parents and health care professionals support the program.

Trial Registration: Dutch Trial Registration: NTR4808; www.trialregister.nl (Archived by WebCite at http://www.webcitation.org/6cfAYHcYb)

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KEYWORDS

child; chronic kidney failure; family health; health promotion; intervention mapping; parents; program development; telemedicine

Introduction

Parents Managing Their Child's Chronic Disease

Pediatric chronic diseases affect the life of many children, as well as their families [1]. Parents have a key role in managing their child's disease [2,3]: they have to balance their child's health care needs against those of other family members and work commitments [2]. The increased strain on the parenting role, hospitalizations, child function impairments, and difficulties in accepting their child's disease cause emotional problems and high levels of stress among these parents [2,4,5]. This leads to negative effects on their child's health outcomes [1,3,5] and their quality of life [6,7].

Chronic kidney diseases (CKD) are an example of pediatric chronic diseases, posing a lot of psychological tensions on parents, as well as the children themselves [8]. Children with CKD are generally diagnosed early in life, and poor growth and development are frequently seen [9,10]. Infections, bone disease, reduced renal function, and eventually kidney failure are frequent complications among these children [11,12]. Despite interventions like renal replacement therapy or kidney transplantation, mortality remains 30 times higher compared to healthy children [11].

The care for children with CKD is complex for parents, due to complicated medication schedules, nutritional restrictions, and invasive procedures such as three times weekly hemodialysis or daily nocturnal peritoneal dialysis [12]. The parents become nurses, pharmacists, and physicians in addition to their usual parental responsibilities [12]. To help these parents cope with difficulties encountered during all stages of their child's CKD, support and information are necessary [12]. However, interventions to assist these parents with the day-to-day management of their child's CKD and its consequences are lacking.

Online Support Programs for Parents

In 2008, Swallow et al [13] described the need among parents of children with CKD for continuously available, accessible, and reliable support. Online support programs are readily accessible and can lead to improvement in users' knowledge, self-efficacy, social support, health behaviors, and clinical outcomes [14-16]. It is not remarkable that the use of online support programs for parents of chronically ill children is increasing [17-20]. Eccleston et al [1] describe in their extensively conducted Cochrane review in 2012 that many (online) support programs for parents of children with asthma, diabetes mellitus, cancer, and skin diseases improve self-efficacy, family functioning, and psychosocial well-being of these parents. However, programs for parents of children with CKD were not included.

In 2014, Swallow et al developed the first online program for parents of children with CKD stage 3-5 in the United Kingdom [21]. In their feasibility study, they concluded that the program has the potential to beneficially affect the parent's perceived competence to manage home-based clinical care for their children [22]. In the Netherlands, we set out to develop an online support program for parents of children with CKD as well, not only focusing on children with CKD stage 3-5, but also taking children with CKD stage 1-2 into account. By developing and providing such an intervention, we aim to improve the child's quality of life.

Aim of the Study

The aim of this paper is to describe the systematic development of an online support program for parents of children with CKD, and how it will be tested.

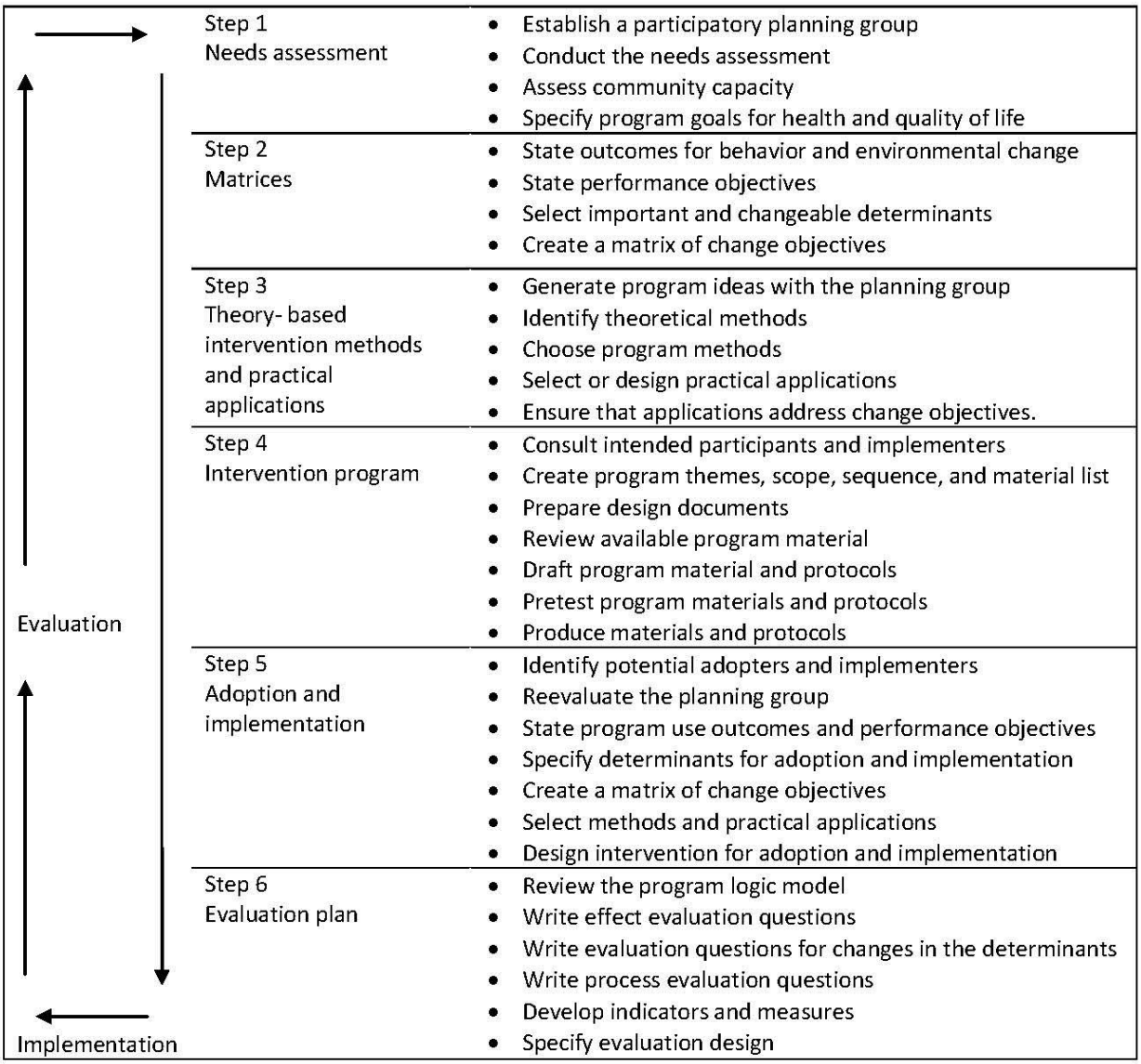
Methods

Intervention Mapping

For the development of the online support program *"e-Powered Parents"*, Intervention Mapping (IM) was used. IM is a protocol for the systematic development of theory- and evidence-based health promotion interventions [23]. It provides health promotion planners with a framework for effective decision making for intervention planning, implementation, and evaluation. It also provides a common creative framework facilitating collaboration between researchers, health promoters, target groups, communities, intermediates, and stakeholders from different backgrounds [23]. IM has already been used for the development of online programs for preventing cyber bullying [24] or stimulating healthy nutrition and physical activity in adolescents [25,26]. Additionally, IM was used to develop support programs for parents of chronically ill children, for example, cystic fibrosis [27].

IM comprises six steps with corresponding tasks (Figure 1 shows this): (1) conducting a needs assessment; (2) identifying intervention outcomes, performance objectives, and change objectives; (3) selecting theory-based methods and practical applications; (4) developing the intervention; (5) planning for adoption and implementation; and (6) planning for evaluating the intervention. These steps will be explained and described in more detail below.

Figure 1. The Intervention Mapping protocol: overview of the six steps and corresponding tasks [23].



Step 1 Conducting a Needs Assessment

The aim of the first step is to conduct a needs assessment to assess the health problem, its related behavior, and the environmental conditions and their associated determinants for the at-risk population [28].

First, a participatory planning group was established, consisting of important stakeholders, potential program users, and implementers. Our planning group consisted of four parents of children with CKD, six health care professionals, and two researchers (WG and BvG). The four unrelated parents were two fathers and two mothers of children with different CKDs in different stadia (for example with chronic kidney failure, on dialysis, and after transplantation). The six health care professionals were members of the pediatric nephrology team: a pediatric nephrologist (EC), a nurse practitioner (JK), a psychologist, a social worker, a dietician, and an educational worker. These professionals were involved in the daily care for children with CKD in a university hospital in the Netherlands.

The participatory planning group helped us to ensure that the intervention addresses issues important to the parents and health care professionals. In every IM step, there was a meeting with the participatory planning group to share knowledge, visions, and experiences aiming to reach consensus and to develop the intervention together.

Second, a needs assessment was conducted. The objective of a needs assessment is to get a clear understanding of the health problem, problem causing factors, and related psycho-social correlates [23].

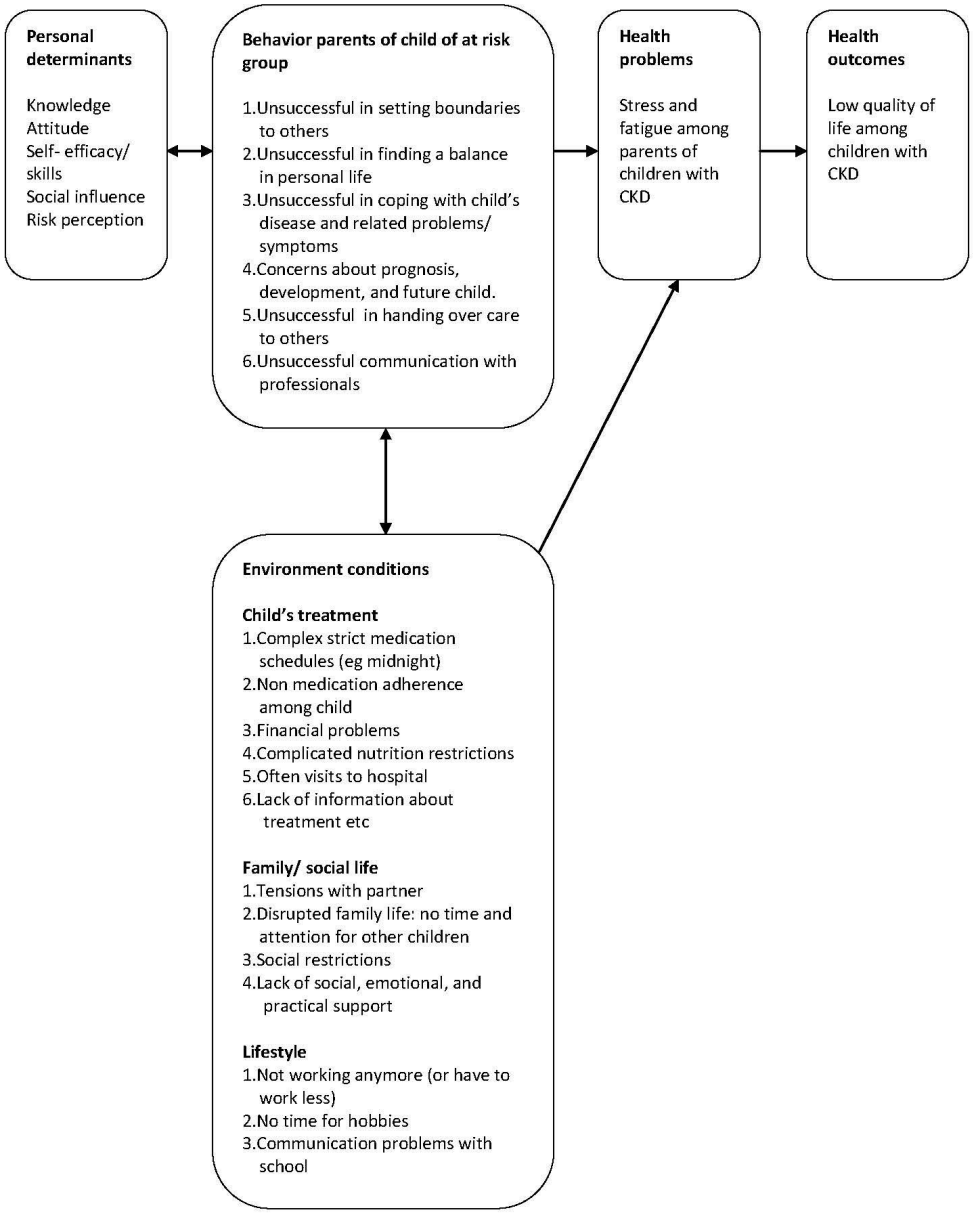
To explore the experienced problems by parents of children with CKD, a literature and focus group study were conducted. In the literature study, PubMed, CINAHL, and PsycINFO were searched for publications between 2003-2013, on experienced problems and support needs expressed by parents in managing

their child's CKD. In the focus group study, five focus group discussions were conducted with parents of children: (1) with hereditary kidney disease (CKD stadium I); (2) with nephrotic syndrome (CKD stadium I); (3) with chronic kidney failure (CKD stadium II-IV); (4) using dialysis (CKD stadium V); and (5) after renal transplantation. All children were treated at the pediatric unit of a university hospital.

The PRECEDE model by Green and Kreuter [29] is used in IM to conceptualize and guide this needs assessment, and to describe the cause of health-related and quality of life problems (Figure 2 shows this). The literature and focus group study both showed that many parents of children with CKD are tired and experience high levels of stress. Parents experience problems regarding their child's treatment (complicated nutritional restrictions, strict medication schedules, and medication nonadherence in their child), their family and social life (tensions with partner, disrupted family life and social restrictions), and their own lifestyle (not being able to work anymore). Additionally, parents experience difficulties in setting their limits, balancing their personal life, and handing over the care for the child to others to create more time for themselves. Often, these problems are influenced by a lack of knowledge, lack of skills, and lack of (emotional, practical, and social) support from peers, family, and health care professionals. The results of the focus group study will be published in a separate article (*manuscript is under review*).

The experienced problems and needs described in the needs assessment were discussed and assessed for completeness with the parents and professionals of the participatory planning group at the end of step 1.

Figure 2. PRECEDE model [23]. Chronic kidney diseases: CKD.



Step 2 Identifying Intervention Outcomes, Performance Objectives, and Change Objectives

The purpose of the second step of IM is to define, as specifically as possible, what should change in the target group and the environment in order to deal with or reduce the health-related problem.

First, program outcomes were stated, based on the needs assessment in step 1 (see Figure 2). Program outcomes are defined as the desired changes in the behavior and the environmental conditions [30]. Program outcomes regarding the behavior of the parents covered managing and coping with their own symptoms, the treatment of their child, their social activities, and their lifestyle. An example of a behavioral program outcome was, “Parents find a balance in their responsibilities as a caregiver for their child, and their own personal life”.

Second, the program outcomes were subdivided into performance objectives. Performance objectives are the required actions to accomplish the change in the behavioral and environmental outcomes [30]. What do parents have to do to find a balance in their responsibilities as a caregiver for their child, and their own personal life? Examples of performance objectives related to this outcome were, “Parents set their limits to others”, and, “Parents hand over their child’s care to others” (see Table 1). The participatory planning group worked together to formulate the performance objectives per program outcome.

Third, determinants were selected per performance objective (see Table 1). Determinants are those factors that are associated with the performance of behavior [30]. The focus was mainly on the determinants attitude, social influence (subjective norms), and self-efficacy (perceived behavioral control) (see Table 1) as the Theory of Planned Behavior (TPB) determined these constructs as the most important determinants of behavior [31]. The TPB has successfully been applied to many types of health behavior [23], and Internet interventions based on the TPB tend to have a large effect on behavior [32]. Besides these three determinants (attitude, social influence, and self-efficacy), knowledge and risk awareness were taken into account as well, as these are necessary prerequisites for these determinants [23].

Parents and professionals of the participatory planning group were asked to rank the determinants per performance objective, taking the importance and changeability of the determinants into account.

Fourth, change objectives were specified, by crossing the performance objectives with their determinants. Change objectives are specific goals stating what should change at an individual level [30]. For example, that what is necessary for parents to learn to hand over their child’s care to others. A change objective for the determinant “knowledge” is thereby, “Parents express the consequences of their daily care for their child and not handing over the care, on symptoms such as stress and fatigue” and a change objective for the determinant “attitude” is, “Parents express the benefits of handing over their care for their child to others”. For more change objectives, see Table 1.

Table 1. Examples of performance and change objectives of the behavioral outcome “Parents find a balance in their responsibilities as a caregiver for their child, and their own personal life.”

Performance objectives	Change objectives				
	Knowledge	Attitude	Self-efficacy	Skills	Social influence
Parents set their limits to others	Can mention ways to set their limits and say no	Express the benefits of setting their limits	Express their confidence in setting their limits and saying no to others, without regret	Describe step by step how they will say no to others and set their limits	Recognize that their social environment may be unaware of the burden and problems they experience
	Explain the relation between stress and fatigue and not setting their limits	Express positive attitudes toward setting limits and saying no			
Parents hand over their child’s care to others	Express the consequences of their daily care for their child and not handing over the care, on symptoms such as stress and fatigue	Express the benefits of handing over their care for their child to others	Express their confidence to hand over their care	Describe how they will hand over their care	Recognize that their environment really wants to support them and will take over the care
		Express positive attitude toward handing over their care for having more time for themselves			
		Accept the possibility that the caregiver can make mistakes			

Step 3 Selecting Theory-Based Methods and Practical Applications

The aim of the third step is to review theoretical methods to effect changes in the health behavior of the individual and to change organizational and societal factors to affect the environment [33].

First, theoretical and empirical literature were searched for theory-based methods. Methods have their origins in behavioral and social science theories and are general techniques or processes for influencing changes in determinants of behavior and environmental conditions (see step 2) [33]. For example, to reach the change objective "Parents express the benefits in handing over their care for their child to others", it is important to look at methods for changing the determinant "attitude."

Second, for the selection of these methods, the Basic Methods for Behavioral Change described by Bartholomew et al [23] and the Coding manual for behavioral change techniques (BCTs) [34] were used. BCTs are comparable to Basic Methods for Behavior Change and based on the taxonomy of Abraham and Michie [35]. BCTs to change the determinant attitude are for example "Reevaluation of outcomes" and "Persuasive communication".

There were two researchers (WG and BvG) who independently chose the BCTs per change objective and discussed them together until consensus was reached. The most often selected BCTs were "Providing general information" (knowledge), "Self monitoring of behavior" (awareness), "Persuasive communication" (attitude), "Reevaluation of outcomes" (attitude), "Practice" (self-efficacy), "Modeling" (self-efficacy), "Information about peer behavior" (social influence), and "Use of social support" (intention). Most often, a combination of BCTs was chosen, because this is most effective in promoting behavior change [32,36,37].

Third, the chosen methods were translated into practical applications (see Table 2). A practical application is a specific technique for the practical use of theoretical methods in ways that fit with the target group and the context in which the intervention will be conducted [23]. Examples of applications for the method modeling include role playing activities or videotaped role models. In selecting the practical application, the parameters were taken into account. Parameters are conditions under which the theoretical method will be effective. A parameter of modeling, for example, is that the individual can identify him or herself with the model [23].

The final selection of the methods and practical applications was decided in a meeting with the four parents and individual meetings with the health care professionals of the participatory planning group.

It turned out that regarding the "environmental conditions" (see Figure 2), it was quite impossible to change the problems in an online program. Therefore, the decision was made to give the parents information and teach them how to cope with these environmental problems (see step 4).

Table 2. An overview of the determinants, used methods, and parameters in the training module "Setting limits".

Determinant	Method (and related theory)	Parameters for use
Knowledge	Advanced organizers	Schematic representations of the content or guides to what is to be learned.
	Elaboration	Individual with high motivation and cognitive ability; messages that are personally relevant, surprising, repeated, self-pacing, not distracting, easily understandable; messages that are not too discrepant and cause anticipation of interaction.
Awareness	Self monitoring of behavior	The monitoring must be of the specific behavior (that is, not of a physiological state or health outcome). The data must be interpreted and used. The reward must be reinforcing to the individual.
	Self reevaluation/ consciousness raising	Can use feedback and confrontation; however, raising awareness must be quickly followed by increase in problem solving ability and self-efficacy.
Attitude	Persuasive communication	Messages need to be relevant and not too discrepant from the beliefs of the individual; can be stimulated by surprise and repetition. Will include arguments. For central processing of arguments they need to be new to the message receiver.
Social influence	Provide information about peer behavior	Positive expectations are available in the environment.
	Stimulate communication to mobilize social support	Combines caring trust, openness, and acceptance with support for behavioral change; assumes that positive support is available in the environment.
Self-efficacy	Planning coping responses	Identification of high risk situations and practice of coping response.
Skills	Guided practice	Sub skill demonstration, instruction, and enactment with individual feedback; requires supervision by an experienced person; some environmental changes cannot be rehearsed.
	Modeling	Attention, remembrance, self-efficacy, and skills, reinforcement of model, identification with model, coping model instead of mastery model.
	Feedback	Feedback needs to be individual, follow the behavior in time and be specific.

Step 4 Developing the Intervention

In this fourth step, the aim is to combine the chosen applications of step 3 into a program and to develop working documents to guide the program production [38].

The components of the program were developed with the parents and health care professionals of the participatory planning group. For example, for the performance and change objectives regarding "Setting limits", parents were asked to write a testimony regarding how they set their limits and the problems they experience. The information and materials were discussed with the social worker and psychologist.

The information technology company transformed the information into an online program, with *e-Powered Parents* as a result (the Dutch title of the online program is *"Mijn Kinderniernet"*).

e-Powered Parents comprises three components: (1) an informative part, (2) an interactive part, and (3) a training platform consisting of four training modules.

The informative part comprises information about different kidney diseases, treatment possibilities (including medication and nutritional restrictions), and (financial) regulations. Additional to the information, there are several folders and videos for parents and their children. This informative part focuses mainly on the environmental problems (see Figure 2).

The interactive part consists of a chat room where parents are able to chat with peers and health care professionals. Additionally, there is a forum where parents can ask questions and share their experiences, tips, and tricks with peers. Parents also have the opportunity to send private messages to other parents.

The training platform consists of four different training modules, based on the problem related behavior of the parents (see Figure 2): (1) "Managing stress"; (2) "Setting limits" (see Table 3); (3) "Communicating"; and (4) "Coping with your child's CKD". Every training module consists of several sessions (minimal two and maximal five). Each session starts with a welcome page, followed by a short introduction explaining what parents can expect to learn in this session, and what they have learned in the previous session. The training modules are not obligatory and can be saved temporarily. Parents themselves can select the training modules and conduct them in no particular order and as often as they want.

The program was pretested in the planning group; parents and health care professionals were asked to look closely at the information and testimonies, exercises, the ease of use, comprehensibility, and lay out of the training modules. The program was adapted using their feedback: testimonies were, for example, redrafted and spelling errors removed.

Table 3. Topics and sessions in the online training module "Setting limits".

Session	Topics
1. Welcome	• Short introduction in why setting limits is important for parents and what they will learn in this training module • Testimony by parent why it is hard to set your limits • Information how they can ask their social environment for support
2. Saying "no", why it is important	• Test how easily parents say no • Information about why it is important to say no, the advantages, and why it is so difficult • Testimony by parent why it is difficult • Information about thoughts and their influence on saying no • Exercise to write down their thoughts
3. Saying "no", how to do?	• Information about different ways to say no (sub assertive, assertive, aggressive), steps in how you can say no and what is important • Exercises to say no, varying from easy to difficult and to become aware what went right and wrong • Tips to discuss the exercise with their partner or friends
4. Handing over the care	• Testimonies of parents why it is difficult to hand over the care for their child to others • Information about why it is important to hand over their child's care • Exercise to hand over their child, for example: to become aware of the advantages of handing over care • Exercise with their partner to discover which activities they find important and how they can make time for it • Exercise to define their social network, to discover who can provide what kind of support • Tips by health care professionals and parents to hand over care • Exercise to describe what they can do when things go wrong • Phone numbers of health care professionals when parents find it hard to hand over their care

Step 5 Planning for Adoption and Implementation

The aim of the fifth step is to design a plan for the diffusion and delivery of *e-Powered Parents*. From the start of the planning process, implementation is anticipated, thereby involving parents and professionals of the participatory planning group [39].

First of all, health care professionals who are involved in the adoption and implementation of *e-Powered Parents* were identified. The key implementers, the nurse practitioner (JK) and the pediatric nephrologists (MC), were part of the participatory planning group and were involved in the development of our program. Additionally, in the weekly multidisciplinary meetings at the pediatric nephrology unit, the nurse practitioner informed the health care professionals about

the progress of *e-Powered Parents*. Furthermore, a guideline with instructions was developed for parents and health care professionals for using *e-Powered Parents*.

To stimulate parents to use *e-Powered Parents*, several practical applications are planned in advance: parents who do not log in will be reminded of *e-Powered Parents* by email and during their consult with the health care professionals in the hospital. Parents who do log in will receive an email regularly with news items and updates on *e-Powered Parents*. Examples of news items are recipes for parents during birthday parties, information about yearly changes in financial regulations, or invitations for meetings. Additionally, parents will be invited to ask their questions on the forum, where health care professionals and peers are able to respond.

Step 6 Planning for Evaluating the Intervention

The objective of the final step of IM is to design an evaluation plan specifying which information needs to be collected from the parents to have insight into the effect of the program [40].

The effect evaluation will be determined in a feasibility study, using an explorative randomized control trial (RCT) (Dutch Trial Register: NTR4808). The Medical Research Counsel guidance states that feasibility studies are essential in the development and testing of an intervention prior to a large scale evaluation [41].

The aims of the feasibility study will be to: (1) identify outcome measures most likely to capture potential benefit; (2) evaluate the potential effectiveness and effect size of *e-Powered Parents*; and (3) evaluate continued use or dropping out of *e-Powered Parents*.

Parents of 120 families, including those (1) with hereditary kidney disease (CKD stadium I); (2) with nephrotic syndrome (CKD stadium I); (3) with chronic kidney failure (CKD stadium II-IV); (4) using dialysis (CKD stadium V); and (5) after renal transplantation will be included in the explorative RCT. Stratified randomization (at family level) will be used to allocate equal numbers of parents in each of the five different categories in the control and intervention group. Parents in the control group receive the usual care, consisting of regular care and treatment for their child at a university hospital in the Netherlands. Parents in the intervention group additionally have the opportunity to use *e-Powered Parents* for six months. Both parents of a child can participate in the study, and will be randomized together into the control group or intervention group.

To explore which outcome measures are most likely to capture potential benefit, and to evaluate potential effectiveness, five outcomes were chosen: (1) The *child's quality of life* will be measured using the Child Vulnerability Scale [42]. This proxy instrument measures the parents perceived child vulnerability, which is related to the child's health-related quality of life [43]. (2) *Parental stress* will be measured using the Pediatric Inventory for Parents [44]; and (3) *Parental fatigue* using the Multidimensional Fatigue Inventory [45]. (4) *Self-efficacy in the communication with health care professionals* will be measured using the Perceived Efficacy in Patient-Physician Interactions [46], and (5) *Family management* using the Family Management Measure [47].

The selection of these five main outcomes is based on the PRECEDE model in the first step of IM (see Figure 2). The child's quality of life is the "Health outcome". Parental stress and fatigue are the "Health problems" among parents. Self-efficacy in the communication with health care professionals is a "Behavior problem". Family management is an overall outcome, indicating the experienced difficulties by parents in managing the condition of their child.

The data will be collected, using online questionnaires, at baseline (T0) and after six months, at the end of the intervention period (T1).

For the first aim (to identify outcome measures most likely to capture potential benefit), commonly used indicators to determine the sensitivity of the outcome measures will be used; for example, floor and ceiling effects, percentage of subjects showing no change in score between T0 and T1, and the effect size of the change score [48].

For the second aim (to evaluate the potential effectiveness and effect size of *e-Powered Parents*), multilevel analysis will be used to test for the posttest differences on the five outcome measures between the intervention and control group. In the multilevel analysis, there will be adjustments for families consisting of one, two, three or even four parents, and also for parents with two or more children with CKD.

The third aim (to evaluate continued participation or dropping out) is part of the process evaluation. For the process evaluation, the framework of Linnan and Steckler [49] will be used, which is endorsed by Bartholomew [23]. The key components that will be taken into account in the process evaluation are the context, reach, dose delivered, dose received, fidelity, and recruitment of the intervention [49]. Different kinds of data will be used. First, data from *e-Powered Parents* will be extracted to answer questions such as "how often did parents log in" and "which components did they visit?" Second, a short extra questionnaire will be used in the T1, to explore parents' experiences on, for example, the ease of use of the program. Third, interviews will be conducted with parents to explore their experiences with *e-Powered Parents*. Parents who regularly visit the program will be asked about their experiences (positive and negative), their ideas to improve the program, and in which way and how the program supported parents in their stress management, setting limits, communicating with health care professionals, and coping with their child's disease. Parents who did not log in (drop out), or only logged in once, will be asked about their reasons for not logging in and possible barriers they experienced. Additionally, interviews with health care professionals will be conducted to explore their experiences and their views on the adaptation of the program and their own role.

Results

Study results are expected to be published in the summer of 2016.

Discussion

Principal Findings

The aim of this study was to describe the systematic development of an online support program for parents of children with CKD, to reduce parental stress, and thereby improve their child's quality of life.

In the development of this online support program for parents we used IM. IM has been a useful protocol for several reasons. First, it provided us with a framework to structure the development process. It enabled us to systematically use theory, empirical evidence, and practical perspectives in the development of the intervention. Second, the formation of the multidisciplinary planning group consisting of parents, health care professionals, and researchers was valuable. Knowledge, experiences, and visions of the different members were shared. Using the participatory planning group in the developmental process, we were ensured that parents and health care professionals support this program. Third, the needs assessment in step 1 was very helpful in exploring and understanding the problems parents of a child with CKD are facing and the needs for support they have. It helped us to ensure the program addressed the needs of these parents. Fourth, although creating the matrices of the performance and change objective in step 2 was very time consuming and might become very extensive in the case of many different behaviors, the matrices were convenient in deciding which behavior the intervention should be targeting. And fifth, the review of theoretical methods in step 3 was very useful in selecting practical applications.

However, there were also challenges. First of all, the target group of this intervention is quite undifferentiated, namely parents of children with different kinds of CKDs and in different stadia. For the four parents in the participatory planning group, there were some difficulties empathizing with parents of children with different kinds of CKD and their experienced problems and needs. Moreover, in contrast to the normal route of IM, we knew already in step 1 that we wanted to develop an online program. Usually, the channel of the program is chosen in step 4. Although we are still convinced that an online program is the best channel for these parents, it might have influenced our choice for relevant methods and applications in step 3. For example, goal setting, an effective method to influence intention, was not applied in *e-Powered Parents*. Interaction between health care professionals and parents is, for example, not possible in the training modules. Subsequently, health care professionals are not able to support parents in setting their (sub) goals. Another challenge is that although the program is systematically and evidence-based developed, its success depends on its use by the parents and professionals in everyday practice.

To our knowledge, only Swallow et al [21,50] have developed an online program for parents of children with a CKD stage 3-5. There are similarities and differences between both programs. Like our program, Swallow's program focuses on clinical care giving information and psychosocial support by using information, videos, family case studies (blogs), chat function, and a question and answer area. Both programs will also focus on managing parental stress. In our program, we also intend to change the behavior of parents in setting their limits, communicating with health care professionals, and coping with their child's disease. Moreover, Swallow et al focus on parents of children with CKD stage 3-5, while we additionally focus on children with CKD stage 1-2. Because of these differences and similarities, it would be very interesting to compare our results after the evaluation.

Parents and health care professionals in the participatory planning group were enthusiastic about the online program and its feasibility. If the explorative trial demonstrates sufficient effectiveness of this online program, this program could be embedded in more university hospitals in the Netherlands.

Conclusions

By applying IM, we were able to create a unique and promising online support program for parents of children with CKD in the Netherlands. Our explorative trial will indicate whether the intervention improve their child's quality of life.

Acknowledgments

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Authors' Contributions

WG drafted the manuscript and BvG, JK, EC, LS, and GK edited the manuscript.

Conflicts of Interest

None declared.

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Abbreviations

BCTs: behavioral change techniques
CKD: chronic kidney disease
IM: Intervention Mapping
RCT: randomized control trial
T0: at baseline data collection
T1: at the end of the intervention period data collection
TPB: Theory of Planned Behavior

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Protocol

Inclusion of Ethnic Minorities in Telehealth Trials for Type 2 Diabetes: Protocol for a Systematic Review Examining Prevalence and Language Issues

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Abstract

Background: Type 2 diabetes is common, on the rise, and disproportionately affects ethnic minority groups. Telehealth interventions may mitigate diabetes-related complications, but might under-recruit or even exclude ethnic minorities, in part because of English language requirements. The under-representation of minority patients in trials could threaten the generalizability of the findings, whereby the patients who might stand to benefit most from such interventions are not being included in their evaluation.

Objective: The aims of this systematic review are twofold: (1) to assess the reporting and prevalence of ethnic minorities in published telehealth trials for type 2 diabetes, including identifying trial features associated with successful patient recruitment; and (2) to determine the proportion of such trials that report English language proficiency as an inclusion/exclusion criterion, including how and why they do so.

Methods: Randomized controlled trials (RCTs) of adults with type 2 diabetes in Western, English-speaking countries that included telehealth interventions targeting diabetes as a primary condition, and those that did not specifically recruit minority groups will be included. Search strategies were devised for indexed and keyword terms capturing type 2 diabetes, telehealth/health technology, and RCTs in English language publications from 2000 to July 2015 in MEDLINE, PsycINFO, EMBASE, CINAHL, and CENTRAL. Reference lists of included studies will also be searched. Two reviewers will independently screen abstracts and full-text articles against inclusion criteria, mediated by a third reviewer if consensus cannot be reached. Data extracted from included studies will be checked by a second reviewer and will be summarized using narrative synthesis.

Results: This research is in progress, with findings expected by Spring 2016.

Conclusions: This review will address research reporting and recruitment practices of ethnic minorities in telehealth RCTs for type 2 diabetes. Prevalence estimates will elucidate generalizability of existing research, with implications for researchers, health professionals, and policy makers. Identifying trial or intervention features that appear to facilitate ethnic minority recruitment, as well as language barriers that impede it might suggest ways to improve recruitment in future trials.

Trial Registration: PROSPERO International Prospective Register of Systematic Reviews: CRD42015024899; http://www.crd.york.ac.uk/PROSPERO/display_record.asp?ID=CRD42015024899 (Archived by WebCite at http://www.webcitation.org/6fUMqbJ0f).

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KEYWORDS

telemedicine; telehealth; type 2 diabetes; diabetes mellitus; ethnic minorities; trial recruitment; external validity; systematic review; English proficiency; health communication

Introduction

Diabetes is a common chronic condition, with global estimates of 387 million diabetic adults worldwide in 2014 [1]. This figure is projected to rise rapidly in the coming years [1], which will further increase the financial burden on health care systems. About 90% of diabetic patients have type 2 diabetes, which typically affects those of middle age or older, although the growing incidence of childhood obesity means that this condition is now occurring in younger populations as well [1]. Risk of developing type 2 diabetes can be reduced by making healthier lifestyle changes, while subsequent type 2 diabetes-related complications, such as heart disease and stroke, can be reduced from good diabetes management [2,3]. Therefore, many behavioral interventions, including those making use of telehealth, which is defined here as the use of technology to support the remote delivery of health care and promote self-management [4], have targeted this condition.

Telehealth could help health systems to cope with the increasing pressures they face with an aging population experiencing rising rates of chronic conditions, and also empower patients to self-manage their care. A key benefit of telehealth is that it can improve access to care for those who experience difficulties utilizing traditional health care [5]. Indeed, telehealth is attracting international interest as an alternative to traditional face-to-face care [6]. While the effectiveness evidence of telehealth from randomized controlled trials (RCTs) is mixed and might vary by chronic condition, systematic reviews of telehealth interventions for type 2 diabetes have generally shown beneficial clinical effects, such as improved blood glucose levels [7-9]. However, many telehealth trials conducted in Western countries recruit mostly white patients [10-12] or may not report the ethnicity of participants [13].

The under-recruitment of certain sociodemographic groups, such as ethnic minority populations, to telehealth RCTs jeopardizes the generalizability of trial findings. This issue is particularly pressing in the case of type 2 diabetes, because the condition is about six times more prevalent in South Asians and up to three times more common in those of African and Afro-Caribbean descent [14]. Across ethnic minorities, the prevalence of type 2 diabetes is 5.7%, whereas it is 1.7% for non-Hispanic white ethnic groups [1]. If the effectiveness and acceptability of telehealth interventions for type 2 diabetes are mostly being trialed among white patients, whether the same interventions would be equally beneficial in ethnic minority groups—those with comparatively greater health needs in this regard—is not being adequately addressed. This has implications when considering new service delivery deployment of such telehealth interventions.

There is some evidence for the low levels of ethnic minority participation in telehealth interventions for diabetes, despite the fact that ethnicity is not widely reported. In a systematic review of 26 studies of interactive computer-based interventions for diabetes, only 8 of these studies reported on the inclusion of ethnic minorities, whereby the median proportion of ethnic minority participation was 39% of the recruited study sample (within-study range of ethnic minority participants: 5-100%) [15]. Further to this initial investigation, a secondary aim of a recent review by Cotter et al [16] was to examine the degree to which Internet interventions were tailored to diverse or underserved diabetic populations. The ethnic mix of participants was reported in 4 of the 9 studies included in this review, whereby there was between 24% and 100% minority group participants across these 4 studies. Another review published in the same year (2014) reported that half of the 16 included RCTs contained minority prevalence information, in which the within-study range of ethnic minority participants was 15-100% [9].

While these reviews provide some indication about the lack of minority patient participation and the under-reporting of ethnicities in telehealth research for diabetes, they are subject to several limitations. These consist of including both RCTs and other study designs within the review [15,16], including studies of patients with type 1 and type 2 diabetes [9,15], and only considering a narrow range of computer technologies, to the exclusion of other telehealth interventions [15,16]. RCTs are held as the “gold standard” in health research because they reduce bias and potential confounding, but they may attract participants who differ from those taking part in other types of studies, due to the time commitment required for participation and other factors [17,18]. Furthermore, since it is only type 2 diabetes that disproportionately affects ethnic minority groups, rather than type 1 diabetes, it is more important to establish the prevalence of ethnic minorities in studies that are restricted to type 2 diabetic patients. In addition, excluding studies that make use of glucose monitoring or that are telephone based means that a large volume of telehealth research in this area was not considered. Finally, a fundamental issue affecting prevalence estimates of ethnic minorities in the 2 more recent reviews [9,16]—albeit not the focus of either review—is that they included studies that, during recruitment, specifically targeted just one or more ethnic minority groups. This is problematic because it overestimates and biases prevalence estimates, as well as masks overall user acceptance in the general patient population. To overcome these limitations, a more comprehensive systematic review of the literature is required, as well as exploring potential barriers and facilitators to ethnic minority participation in telehealth trials.

One frequently cited challenge in reference to minority group participation in RCTs relates to patients’ language proficiency and literacy [19,20]. Among those living in the United States, around 25 million people are unable to speak English fluently [21], while it is estimated that almost 300,000 adults in England and Wales from the 4 most common ethnic minority groups speak little or no English [22]. Ensuring that patients have the requisite language ability to understand the conditions of research participation is essential in all research studies for

ethical reasons (eg, obtaining informed consent). Moreover, because a fundamental component of telehealth interventions involves non-face-to-face communication, being able to engage in efficient communication is integral to participating in and benefiting from telehealth interventions. It is unclear whether researchers are utilizing objective and systematic ways of assessing whether patients have the necessary language skills to participate in telehealth RCTs for diabetes, or whether such decisions are made more subjectively. The role that language plays in telehealth trial participant inclusion or exclusion decisions, therefore, needs to be systematically investigated. This is of particular significance for type 2 diabetes trials, where minority patients who potentially stand to benefit most from such interventions need to be proportionally represented, and language barriers may impede their ability to do so in telehealth trials targeting this condition. To our knowledge, no systematic review has examined language as a potential barrier to ethnic minority participation in telehealth trials for type 2 diabetes.

Taken together, the potential under-recruitment of ethnic minorities and language barriers present two major issues facing telehealth RCTs for type 2 diabetes. Both affect the generalizability of trial findings and have wider implications for the adoption of new telehealth services into health care systems, as well as equitable access to health care. Therefore, the aims of this systematic review are twofold: (1) to assess the reporting and prevalence of ethnic minorities in published telehealth trials for type 2 diabetes, including identifying trial features associated with successful patient recruitment (trial includes ≥30% ethnic minorities, in line with median prevalence across earlier reviews [9,15,16]); and (2) to determine the proportion of such trials that report English language proficiency as an inclusion/exclusion criterion, including how and why they do so. Building on the previously mentioned reviews [9,15,16], our study will update and refine the ethnic minority prevalence figures with more stringent criteria (ie, RCTs only, adult type 2 diabetes patients only, excluding studies with an ethnically targeted sample) intended to minimize the effects of extraneous variables that could bias estimates. Compared with the earlier reviews, we will also include a broader range of telehealth interventions that make use of any technology medium. In addition, we will move beyond simple reporting and seek to identify the characteristics of trials that have higher proportions of ethnic minority participation.

Methods

This systematic review will be conducted in accordance with the *Cochrane Handbook for Systematic Reviews of Interventions* [23] and will follow the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines [24].

Search Strategy

We will search for potentially eligible studies using MEDLINE (via Ovid), PsycINFO (via Ovid), EMBASE (via Ovid), CINAHL (via EBSCOhost), and the Cochrane Central Register of Controlled Trials (CENTRAL, via Wiley Online Library) from January 1, 2000, to July 31, 2015. Reference lists of included studies will also be searched.

Both Medical Subject Heading (MeSH) terms and keywords will be used in the searches to capture themes of type 2 diabetes, telehealth/health technology, and RCTs (see Multimedia Appendix 1 for MEDLINE example). Because of the varied terms researchers tend to use to describe telehealth, the search incorporates numerous plausible key terms. We also examined key terms used in related diabetes telehealth systematic reviews with similar inclusion and exclusion criteria to ensure comprehensive searching [7-9,25]. We consulted a medical subject librarian to finalize the strategy. Search filters restrict results to English language publications, humans, and the relevant publication dates.

Screening and Study Selection

The database searches will be performed by one reviewer (LE). Citations and abstracts will be uploaded into EndNote X7, de-duplicated, and screened for eligibility against the inclusion criteria by the same reviewer (LE). A second reviewer (KB) will concurrently and independently assess the titles and abstracts against the inclusion criteria. Discrepancies will be resolved through discussion and achieving consensus, or by consulting a third reviewer (TI) if consensus cannot be reached. Reasons for excluding abstracts will be recorded for later reporting.

After retrieving the full text of the studies that remain after the first stage of assessment, 2 reviewers (LE and DW) will independently review the full text for further eligibility assessment. Reasons for exclusion will be recorded for reporting in the PRISMA flow diagram. As before, discrepancies will be resolved through consensus or by consulting a third reviewer (TI) when consensus cannot be reached. Multiple reports from the same study will be linked for included studies.

Inclusion Criteria

The following criteria will be used to select studies for inclusion in the review.

Study Designs

Only RCTs will be included, which could include two- or more arm trials, pilot studies, cross-over designs, cluster RCTs, and so on.

Participants

The review will be limited to adult patients (aged ≥ 18 years) of either sex with type 2 diabetes in Western countries where English is both an official and a majority language (USA, Canada, the United Kingdom, Ireland, Australia, and New Zealand). Only participants recruited widely from the general patient population will be included, rather than RCTs that specifically aim to include just one or more ethnic groups. This is because we are interested in assessing the prevalence of ethnic minorities in typical telehealth RCTs, in which the focus is simply on achieving the target sample size. Studies that include a homogeneous ethnic minority sample will artificially inflate the prevalence results. However, trials recruiting from other specific contexts (eg, urban/rural; economically deprived/low-income/medically underserved groups) will be permitted. We acknowledge that there may be a relationship between ethnicity and these other sociodemographic

characteristics, but studies that include such samples are unlikely to result in completely homogeneous ethnic samples. In addition, telehealth is meant to address barriers to accessing traditional health care, and, therefore, recruitment within these contexts is a direct test of a typically cited advantage of telehealth [26].

Interventions

Telehealth interventions of any duration, using any technology medium specifically designed to treat or improve type 2 diabetes as the primary condition, will be included. By this, we mean interventions targeting or monitoring blood glucose levels, diabetes education/knowledge, medication adherence, and diabetes self-care behaviors (eg, foot checking and exercise/diet/weight interventions), whereas mental health interventions and treatment of other secondary diabetes-related complications (eg, foot ulcers, cardiovascular health) will not be included. As outlined in the "Introduction" section and illustrated in the search strategy (see Multimedia Appendix 1), we take a broad definition of telehealth, which includes telemedicine, telemonitoring, teleconsultations, medical informatics, eHealth and mHealth (electronic or mobile health), or other forms of remote health care delivery, treatment, and support. However, the intervention must be described by the authors as telehealth or the majority of the intervention must be delivered electronically, rather than in-person.

Comparators

We will include studies employing any comparison group within an RCT, such as usual care, wait-list control group, or head-to-head trials.

Outcomes

Studies will not be selected on the basis of any reported outcomes. The primary outcomes of the review will include

1. Proportion of RCTs that report on the ethnic/racial composition of trial participants.

2. Overall prevalence of ethnic minorities reported in telehealth trials for patients with type 2 diabetes (between-study median and range).

3. Features of studies that include a higher proportion of ethnic minorities (≥30% of the sample), such as country, recruitment setting (primary care, community, secondary care), telehealth medium (telephone, video, email, etc), resource availability (translation/interpretation), targeting of low-income recipients, tailoring of intervention (to individual needs, cultural group, etc).

4. Proportion of telehealth trials for type 2 diabetes that report English language proficiency (oral/written) or literacy as inclusion/exclusion criteria.

5. Way in which English language proficiency/literacy is operationalized by authors reporting this as inclusion/exclusion criteria.

6. Reason(s) that patients are excluded on language grounds (eg, ethical considerations, ability to participate in intervention, lack of translation/interpretation resources).

Setting and Timing

There are no restrictions on the study setting or length of intervention or follow-up.

Report Characteristics

The review will include RCTs published in English in peer-reviewed journals between 2000 and 2015. The rationale for this 15-year publication restriction is that we are interested in capturing the current state of telehealth RCTs for type 2 diabetes. Health technology has evolved rapidly within this period and telehealth trials have also proliferated in recent years, and so this time frame should provide an up-to-date and sufficient summary of ethnic minority participation and language recruitment barriers in trials.

Exclusion Criteria

Because of the greater risk of bias and potential confounds, study designs other than RCTs (eg, observational studies, surveys), including nonrandomized controlled studies, will be excluded. The following additional exclusions apply: (1) systematic or other literature reviews, letters, commentaries/editorials, study protocols, conference abstracts, or presentations; (2) secondary subgroup analyses of RCTs or RCT-generated data modeling studies; (3) RCTs not published in peer-reviewed journals, such as dissertations and case reports; (4) targeted ethnic minority trials (could be a single ethnic minority sample, or a dedicated comparison between one or more ethnic groups), although the number of such studies and recruited patients of various ethnic backgrounds that would otherwise have met our eligibility criteria will be recorded; (5) mixed (type 1 and type 2) diabetes samples, type 1 diabetes, gestational (pregnancy) diabetes, or diabetes insipidus; (6) studies involving adolescents or children; (7) telehealth interventions directed at and solely experienced by health professionals, even if the study measures the effect of such interventions on type 2 diabetic patients; and (8) telehealth interventions directed solely at diabetes-related complications, such as diabetic retinopathy, hypertension, diabetes distress. Interventions addressing both primary and secondary diabetes issues will, however, be included.

Data Collection

Data extraction forms will be developed for the current review using existing guidelines [23]. The form will contain information about the (1) study details (eg, country, recruitment setting, design, inclusion and exclusion criteria), (2) participant demographics (eg, sample size, age, sex, ethnic mix, baseline diabetes severity), and (3) intervention characteristics (eg, description, duration, frequency, telehealth medium). A complete list of all variables is available through PROSPERO [27]. Outcome data will be abstracted by one reviewer (DH) into Excel, and independently checked by a second reviewer (DW). Disagreements will be resolved by a third reviewer (TI).

The Cochrane risk of bias tool [28] will be used to assess selection, performance, detection, attrition, reporting, and "other" sources of bias. Accordingly, a judgment of "high," "low," or "unclear" bias along with supporting justification will be provided and recorded using Review Manager (RevMan, version 5.3; Copenhagen: The Nordic Cochrane Centre, The

Cochrane Collaboration, 2014). Two reviewers (LE and DH) will assess all studies, which will be checked by another reviewer (DW). As before, disagreements will be resolved through discussion or by consulting a third reviewer (TI).

We anticipate a high degree of missing or unreported ethnicity data in the studies, as well as few details regarding English language inclusion or exclusion criteria. It is, however, beyond the scope of this review to supplement recruitment-related information provided in refereed journal articles by consulting the gray literature or contacting authors for missing information. While we acknowledge this limitation, this is consistent with the overall goal of the review to systematically document what is and is not reported in peer-reviewed trial publications.

Data Synthesis

Proportions of ethnic minorities included across studies (overall, and by specific ethnic group), as well as frequencies of trials reporting English language as inclusion/exclusion criteria, will be presented for individual included studies and aggregated across included studies.

The results will be presented in a narrative synthesis using the synthesis framework developed by Popay and colleagues [29]. The framework sets out the following 4 key elements to narrative synthesis: (1) developing a theory of how the intervention works, why, and for whom; (2) developing a preliminary synthesis of findings of included studies; (3) exploring relationships in the data; and (4) assessing the robustness of the synthesis. In terms of the first element, which focuses on constructing a theory around the intervention's effectiveness, we will re-frame this according to our final research question concerning what features of telehealth trials for type 2 diabetes tend to successfully recruit a sizeable proportion of ethnic groups. We are interested in the kinds of recruitment techniques, studies, or interventions that appear to result in more ethnically balanced telehealth trial samples (≥30% ethnic minority participants). These insights could inform current recruitment practice for targeting ethnically diverse patients in telehealth studies, taking language support and other factors into account. Thus, we will seek to develop a theory around this facet. We will use an inductive approach, in which a theory will be formulated based on themes or patterns that emerge from the data.

As outlined in Popay and colleagues [29], several tools and techniques will be used in processing the data around each of the elements, which could include textual descriptions, groupings and clusters (eg, by telehealth intervention medium, country), tabulation, thematic analysis, concept mapping, and reflecting critically on the synthesis process. In line with the guidance, we will undertake these processes iteratively and will discuss the emerging results with the research team throughout.

An additional, retrospective subgroup analysis (added after PROSPERO registration) will be carried out on trials that specifically targeted ethnic minority groups as part of their recruitment strategy. The aim of this analysis will be to identify trial features or strategies that were employed that potentially resulted in heightened recruitment of these targeted groups. All such trials will have otherwise met the inclusion criteria for the full systematic review, except that recruitment was aimed at one or more ethnic minority groups.

Results

This systematic review is currently underway, with results anticipated by Spring 2016.

Discussion

This systematic review will provide up-to-date prevalence estimates of research-reporting practices and participation rates of ethnic minorities in telehealth trials for type 2 diabetes that make use of a broad range of technologies. The inclusion of ethnic minorities in type 2 diabetes telehealth research is methodologically important to maximize the external validity of findings, in addition to extending the potential benefits of telehealth type 2 diabetes research to a wider cross section of patients. These factors, coupled with the rapid rise of telehealth interventions, make the need to assess language as a potential recruitment barrier for minority participation, as well as highlighting facilitators to recruitment, all the more pressing. Moreover, determining which kinds of trials or interventions tend to attract a higher proportion of ethnic minorities has significance for a variety of stakeholders in the health care system. This could inform trial recruitment strategies, thereby enhancing the ethnic mix within trials, increasing the external validity of findings, while also ensuring a broad spectrum of diabetic patients evaluate and potentially benefit from such telehealth interventions. From a more macro level, greater insight into the characteristics of trials that successfully recruit a heterogeneous population is synchronous with the overarching goal of promoting greater social inclusiveness and more equitable access to health care.

Ethnic minority participation in type 2 diabetes telehealth RCTs will impact health systems when considering commissioning new services for patients, in which effectiveness across ethnically diverse groups and, in particular, among ethnic minorities that are disproportionately affected by type 2 diabetes must be demonstrated. While the 3 previously cited reviews [9,15,16] marked an important first step in tabulating the prevalence of ethnic minorities included in telehealth interventions for diabetes, they also highlighted how few studies (31-50%) actually reported on the ethnic mix of the study sample. If this is still the case in this up-to-date comprehensive review, despite the widespread adoption of best reporting practices for studies, such as the Consolidated Standards of Reporting Trials 2010 (CONSORT) statement [30], then this suggests the need for further attention in research reporting. These guidelines state that baseline characteristics of trial participants should be reported, which is predicated by the fact that this information is gathered as part of data collection. We contend that this is especially pressing in trials of chronic conditions in which there is known to be variation in prevalence by sociodemographic variables, and that it is precisely these variables that constitute which key baseline characteristics ought to be reported.

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Authors' Contributions

LE and TI jointly conceived and designed this study. LE is first reviewer for abstract and full-text screening, contributed to risk of bias assessment, and wrote the first draft of this paper. TI is the principal investigator, who initially conceptualized and provided overall guidance on this work. LR contributed to the design of an earlier related project. All authors critiqued and provided intellectual input to this manuscript.

Conflicts of Interest

None declared.

Multimedia Appendix 1

MEDLINE search strategy using Ovid interface.

[PDF File (Adobe PDF File), 85KB - resprot_v5i1e43_app1.pdf]

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Abbreviations

CONSORT: Consolidated Standards of Reporting Trials
MeSH: Medical Subject Heading
PRISMA: Preferred Reporting Items for Systematic Reviews and Meta-Analyses
RCTs: randomized controlled trials

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Protocol

Social Media for e-Government in the Public Health Sector: Protocol for a Systematic Review

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Abstract

Background: Public sector organizations worldwide are engaging with social media as part of a growing e-government agenda. These include government departments of health, public health agencies, and state-funded health care and research organizations. Although examples of social media in health have been described in the literature, little is known about their overall scope or how they are achieving the objectives of e-government. A systematic literature review is underway to capture and synthesize existing evidence on the adoption, use, and impacts of social media in the public health sector. A series of parallel scoping exercises has taken place to examine (1) relevant existing systematic reviews, to assess their focus, breadth, and fit with our review topic, (2) existing concepts related to e-government, public health, and the public health sector, to assess how semantic complexity might influence the review process, and (3) the results of pilot searches, to examine the fit of social media within the e-government and health literatures. The methods and observations of the scoping exercises are reported in this protocol, alongside the methods and interim results for the systematic review itself.

Objective: The systematic review has three main objectives: To capture the corpus of published studies on the uses of social media by public health organizations; to classify the objectives for which social media have been deployed in these contexts and the methods used; and to analyze and synthesize evidence of the uptake, use, and impacts of social media on various outcomes.

Methods: A set of scoping exercises were undertaken, to inform the search strategy and analytic framework. Searches have been carried out in MEDLINE, the Cochrane Library, Web of Science, and the Scopus international electronic databases, and appropriate gray literature sources. Articles published between January 1, 2004, and July 12, 2015, were included. There was no restriction by language. One reviewer (AT) has independently screened citations generated by the search terms and is extracting data from the selected articles. A second author (CP) is cross-checking the outputs to ensure the fit of selected articles with the inclusion criteria and appropriate data extraction. A PRISMA flow diagram will be created, to track the study selection process and ensure transparency and replicability of the review.

Results: Scoping work revealed that the literature on social media for e-government in the public health sector is complicated by heterogeneous terminologies and concepts, although studies at the intersection of these three topics exist. Not all types of e-government are evident in the health care literature. Interim results suggest that most relevant articles focus on usage alone.

Conclusions: Public health organizations may be taking it for granted that social media deliver benefits, rather than attempting to evaluate their adoption or impacts. Published taxonomies of e-government hold promise for organizing and interpreting the review results. The systematic review is underway and completion is expected in the beginning of 2016.

Trial Registration: PROSPERO International Prospective Register of Systematic Reviews: CRD42015024731; http://www.crd.york.ac.uk/PROSPERO/display_record.asp?ID=CRD42015024731 (Archived by WebCite at http://www.webcitation.org/6dV1Cin91).

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KEYWORDS

e-government; eHealth; health organizations; social media

Introduction

e-Government, Social Media, and the Health Sector

Governments around the world are being challenged by increasing public demand for institutional transparency, involvement in decision making, and easier access to services, as well as by the financial imperative for greater efficiencies in the business of government itself [1]. To respond to these challenges, they have sought to harness the Internet and related information and communications technologies for sharing information and enabling transactions between governmental bodies, businesses, and citizens, as part of a broader "e-government" agenda [1,2]. One channel through which this is being approached is social media, including networking and dissemination platforms, such as Facebook and Twitter, and bespoke online tools for eliciting feedback and other e-government objectives [3,4]. According to a recent United Nations e-government survey, the use of social media by governments tripled from 2010 to 2012, and rose by another 50% in 2014 alone [1]. Because the public health sector represents a major area of government expenditure for most countries, and while the general literature on social media for e-government may contain transferrable insights, it is important to examine how the use of social media for e-government has been approached in this particular context.

Defining and Classifying e-Government

Although e-government (or eGovernment) is often used as a generic label for digitally mediated government, there is some variation in the use of this term. For example, it has been described to as "analogous to e-commerce, which allows businesses to transact with each other more efficiently and brings customers closer to businesses" [2] and as a means to transform governments' relationships with citizens (government-to-citizen and/or citizen-to-government), employees (government-to-employees and/or employees-to-government), nonprofit organizations (government-to-nonprofit and/or nonprofit-to-government), businesses (government-to-business and/or nonprofit-to-business), and other arms of government (government-to-government) [2,5,6]. The information flows depicted in parentheses above are derived from a taxonomy originally developed by Fang [7].

In 2010, Linders et al (as cited in [8]) suggested several dimensions related to the objectives and intended outcomes underlying the use of social media and how these might affect the work of governmental agencies. These are as follows:

- "Democratic participation and engagement, through which social media technologies are used to involve the public in government decision processes, to foster participatory dialog and policy development and implementation.
- Co-production, through which governments and the public jointly develop, design, and deliver government services to improve service quality, delivery, and responsiveness.
- Crowdsourcing solutions and innovations, whereby governments seek public knowledge and talent to develop innovative solutions to large-scale societal issues.
- Transparency and accountability, through which government is open and transparent regarding its operations to build trust and foster accountability" [8].

Although variants of these taxonomies exist (eg, Linders [9] breaks down his co-production theme into "citizen sourcing," "government as a platform," and "do-it-yourself government"), these two broad frameworks [8,9] are useful for interpreting the results of our review and will be taken into account during this process.

Concepts such as "open government" and "e-governance" also overlap with e-government in important ways, such as through a shared objective for transparency [10]; however, each has somewhat unique connotations and communities of practice. For example, the former emphasizes online public access to government documents and statistics, whereas the latter emphasizes legal and regulatory requirements for effective digital services [11], including e-government [12].

Although there is a body of research describing the adoption of social media by public sector actors such as local authorities [13,14], central and federal governments [6,15], cities [16,17], and municipalities [18], many authors claim that little is known about how these technologies are used by public health organizations [19,20]. Moreover, to the best of our knowledge, no study has specifically investigated the adoption and use of social media by public health organizations, taking the perspective that they are also part of government [21].

e-Government and the Public Health Sector

Determining the scope of the public health sector, in terms of its relationship with concepts of government and e-government, is also challenging. Within medicine, *public health* itself is regarded as a distinct discipline, involving the delivery of population-scale health interventions, such as prevention, screening, wellness, maternal and child health services, surveillance of diseases and risks, and scientific research aimed at understanding and improving the health of populations. By contrast, the *public health sector* is a much larger proposition, encompassing government departments of health, special government agencies tasked with public health activities, health care delivery organizations operating within the public sector, and the networks of voluntary and private-sector organizations that contribute to these activities, also collectively referred to as the *public health system* [22]. Although most countries have an underpinning government health system, the private sector plays a more dominant role in some than in others, due to historical, political, economic, and philosophical differences, which have affected the prioritization of market choice versus social equity [23]. For example, definitions of public health arising from the United Kingdom (operating largely in accordance with the Beveridge Social Insurance Model)

emphasize the role of the government, whereas those from the United States (operating to a Mixed Market-Driven Model) tend to place a greater emphasis on the private sector; however, the World Health Organization's definition encompasses all varieties of health systems, as illustrated by the examples shown in Textbox 1. From the perspective of our review topic, this complexity presents a challenge for bridging concepts of "public health sector" and "e-government," and is likely to affect both the description of relevant studies in the literature and our ability to interpret them within an e-government framework. Nevertheless, governments typically maintain oversight and governance of medicine and health care in most countries, through their legal and regulatory powers, and it may therefore be legitimate to include nongovernmental organizations within the scope of our review. This will depend on the extent to which it is possible to adequately differentiate between governmental and nongovernmental public health activities in the included studies, which have not yet been subjected to detailed analysis or critical appraisal, and the choice will be defended in the main systematic review report.

Textbox 1. Sample definitions of public health/systems from the World Health Organization, the United States, and the United Kingdom.

- World Health Organization: Global policy and surveillance, emphasis on lower income countries

"Public health refers to all organized measures (*whether public or private*) to prevent disease, promote health, and prolong life among the population as a whole. Its activities aim to provide conditions in which people can be healthy and focus on entire populations, not on individual patients or diseases. Thus, *public health is concerned with the total system* and not only the eradication of a particular disease" [24].

- United States: Mixed health care economy, largely private sector. Health care as service

Government (Centers for Disease Control and Prevention, Department of Health and Human Services): "Public health systems include *all public, private, and voluntary* entities that contribute to the delivery of essential public health services within a jurisdiction" [25].

Foundation established by government (CDC Foundation): "Public health is the *science* of protecting and improving the health of families and communities through promotion of healthy lifestyles, research for disease and injury prevention and detection and control of infectious diseases. Overall, public health is concerned with protecting the health of entire populations. These populations can be as small as *a local neighborhood, or as big as an entire country or region of the world*" [26].

- United Kingdom: Predominantly public sector, with optional private services. Health care as universal right

Government (Department of Health): "Public health is about helping people to stay healthy, and protecting them from threats to their health. The government wants everyone to be able to make healthier choices, regardless of their circumstances, and to minimise the risk and impact of illness" [27].

National Professional Society (The UK's Faculty of Public Health): "Public Health is the science and art of promoting and protecting health and well-being, preventing ill-health and prolonging life through the organised efforts of society....Public health is population based, emphasises collective responsibility for health—its protection and disease prevention—*recognises the key role of the state*, linked to a concern for the underlying socio-economic and wider determinants of health, as well as disease, and emphasises partnerships with all those who contribute to the health of the population" [28].

Social Media and Health

The literature on social media in health is large and growing: for example, in 2011, the number of social media-related abstracts in PubMed for the years 2002-2011 was 1471, whereas in 2012, there were already 2,330 returns, according to an analysis by Gholami-Kordkheili et al in 2013 [29], and is likely to be considerably higher today. Industry analysts are regularly charting the uses of social media for health-related purposes, indicating growth trends and their impacts on the health care business sector [30], while academic and policy conferences devoted to understanding the social Web and social media in health are proliferating (eg, [31,32]). Although some studies have clearly described the uses of social media for delivering public health services (eg, [33,34]), or for enabling e-government (eg, [35]), the conceptual links between public health, e-government, and social media have not been well described in the literature.

Formative Scoping

Analysis of Existing Systematic Reviews on Social Media in Health

To gain a better understanding of the relevant concepts, and to establish the need for a new review, we first sought to identify and analyze existing systematic reviews on social media in health. The high-level terms "Social Media," "Systematic Review," and "Health*" were used to search the MEDLINE electronic database. Generated returns (n=27) were then filtered to remove duplicates (n=5), articles found not to be systematic reviews (n=1), and nonrelevant publications (n=1). The relevant systematic or quasi-systematic reviews revealed by this search (n=20) and additional systematic reviews identified through snowballing from the reference lists of these articles or found among the returns generated by our systematic review search query described in [36] (n=16) are presented in Multimedia Appendix 1 (n=36). In each case, the review is summarized in terms of its data sources, context, and focus. In most cases, abstracts were used for this analysis; however, full-text review was also necessary in a few cases where abstracts did not contain sufficient detail to allow a judgment to be made.

The scope of the existing systematic reviews (see Multimedia Appendix 1) varies quite widely. Some aim to synthesize the results of existing interventional studies involving social media, some describe the literature on the uses of social media for purposes such as medical education or professional networking, and others examine the potential of social media for facilitating research through recruitment or secondary analysis of data. However, as can be seen from Multimedia Appendix 1, none

is a comprehensive overview aimed at understanding specifically how social media are adopted and used by health organizations either in the public sector or more widely.

Comparing Social Media in e-Government Generally, and Within the Public Health System

We also wished to compare the concepts used to discuss social media in the e-government literature as a whole, and in the subset of e-government literature focused on health systems, because these represent somewhat different communities of practice. To do this, we analyzed samples of titles and abstracts from each corpus of literature.

For the former, the terms "e-government" OR "eGovernment" OR "government" AND "social media" OR "Facebook" OR "Twitter" OR "YouTube" were applied to the Scopus database, and the titles and abstracts of the 50 most highly cited articles were examined (hereinafter referred to as "generic e-government research"). For the latter, we examined the draft list of about 90 relevant abstracts and titles identified using our search protocol registered in PROSPERO [36] (hereinafter referred to as "health e-government research").

The "Word Frequency" query in the NVivo qualitative analysis software package was used to extract the 35 terms appearing most often within each corpus of titles and abstracts. These are depicted in Figure 1 as "word clouds" representing clusters of related terms, their frequency in the text and their proximity to one another. More than half of the most frequently used words are identical in both bodies of literature, suggesting a considerable degree of overlap, although "government" is a more dominant theme in the former and "health" in the latter.

To gain a more nuanced understanding of the differences and overlaps between the generic and health-related e-governance literatures, we also manually plotted the individual terms according to the corpus of research in which they appeared, as shown in Figure 2.

The results summarized in Figure 2 indicate that, although there is overlap between the two corpuses of research, there are also some noteworthy differences. For example, governmental bodies are commonly referred to as "agencies" in the generic e-government literature and as "departments" and/or "organizations" in the health-related e-government literature. Within the generic e-government literature, concepts such as interaction, transparency, and public participation are prioritized, whereas in the health-related e-government literature, there is a greater emphasis on information sharing and dissemination, as well as engaging with the public to inform care quality improvement and obtain ratings of health services. Words like "using" appear frequently in both corpuses, as do other terms describing actions or high-level objectives such as "engagement," as shown in Figures 1 and 2. This suggests that most of the existing research examining social media for e-government in general, and in the context of health systems, focuses on its use, rather than on its development, implementation processes, or impacts. This may reflect the tendency of government organizations to use off-the-shelf social media tools, reducing the requirement for design, although it may also indicate that organizations are engaging with these technologies in the optimistic belief that they will inevitably deliver benefits, rather than seeking to test this proposition. During our systematic review we will seek to identify formative and evaluative studies of social media as a service in the context of public health e-government.

Another interesting observation was that the explicit terms "e-government or eGovernment" did not appear in the corpus of abstracts on social media in health, when we tried to search for it via NVivo's "Text Search" query, despite the high frequency of the single term "government," although it was explicit in the generic e-government literature. In the next phase of our research, we will analyze the full text of included articles derived from each corpus, to examine whether this finding still holds, as well as to establish the overlaps with the related concepts of "open government" and "e-governance," which we had expected to see independently among the most frequently used terms, at least within the corpus of generic e-government research (although the related term "transparency" appeared).

Figure 1. Words appearing in generic vs. health e-government research.

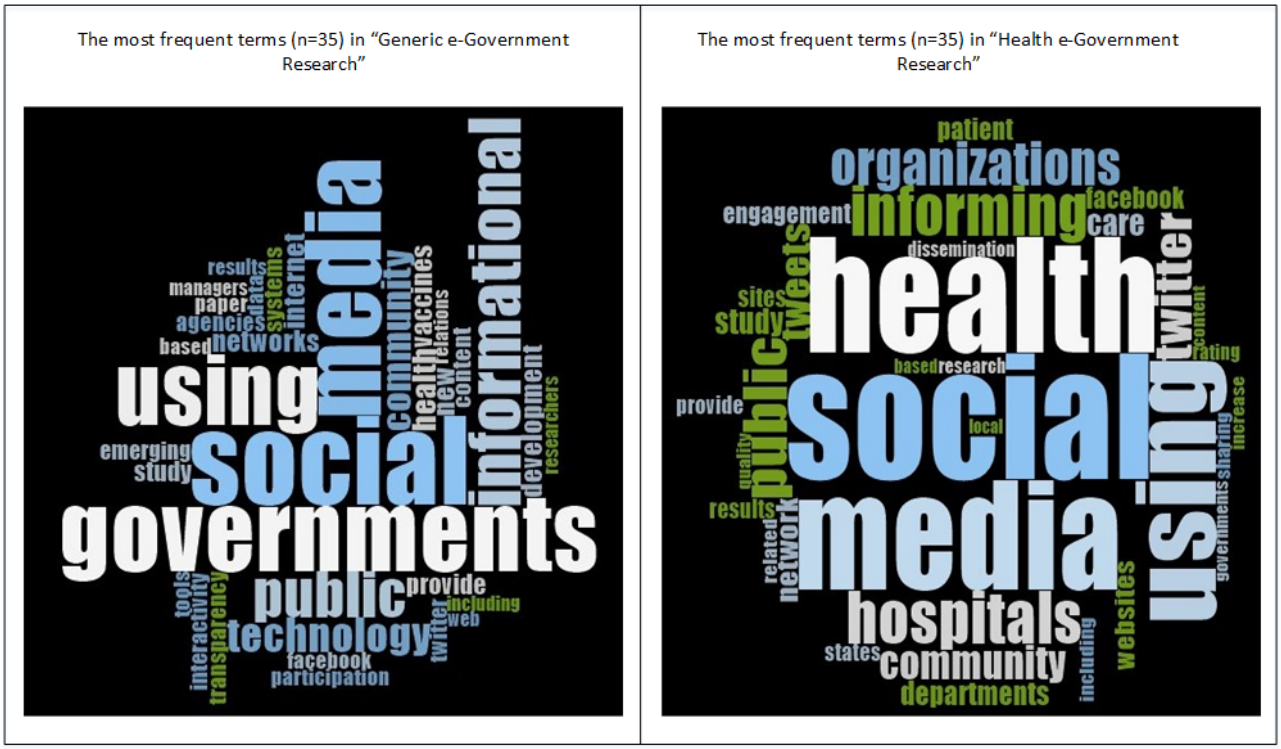


Figure 2. Analysis of terms used in "generic e-government" and "health e-government" research.

Systematic Review Objectives

- To capture the corpus of published studies on the uses of social media by public health agencies or services, at the regional or national levels, in different countries;
- To classify the objectives for which social media have been deployed in these contexts, and the methods used to achieve them, as explicitly stated by the authors or deduced from the published descriptions;
- To analyze and synthesize evidence of the uptake and use of social media by various public sector health organizations and agencies worldwide and their impacts on a range of outcomes.

Methods

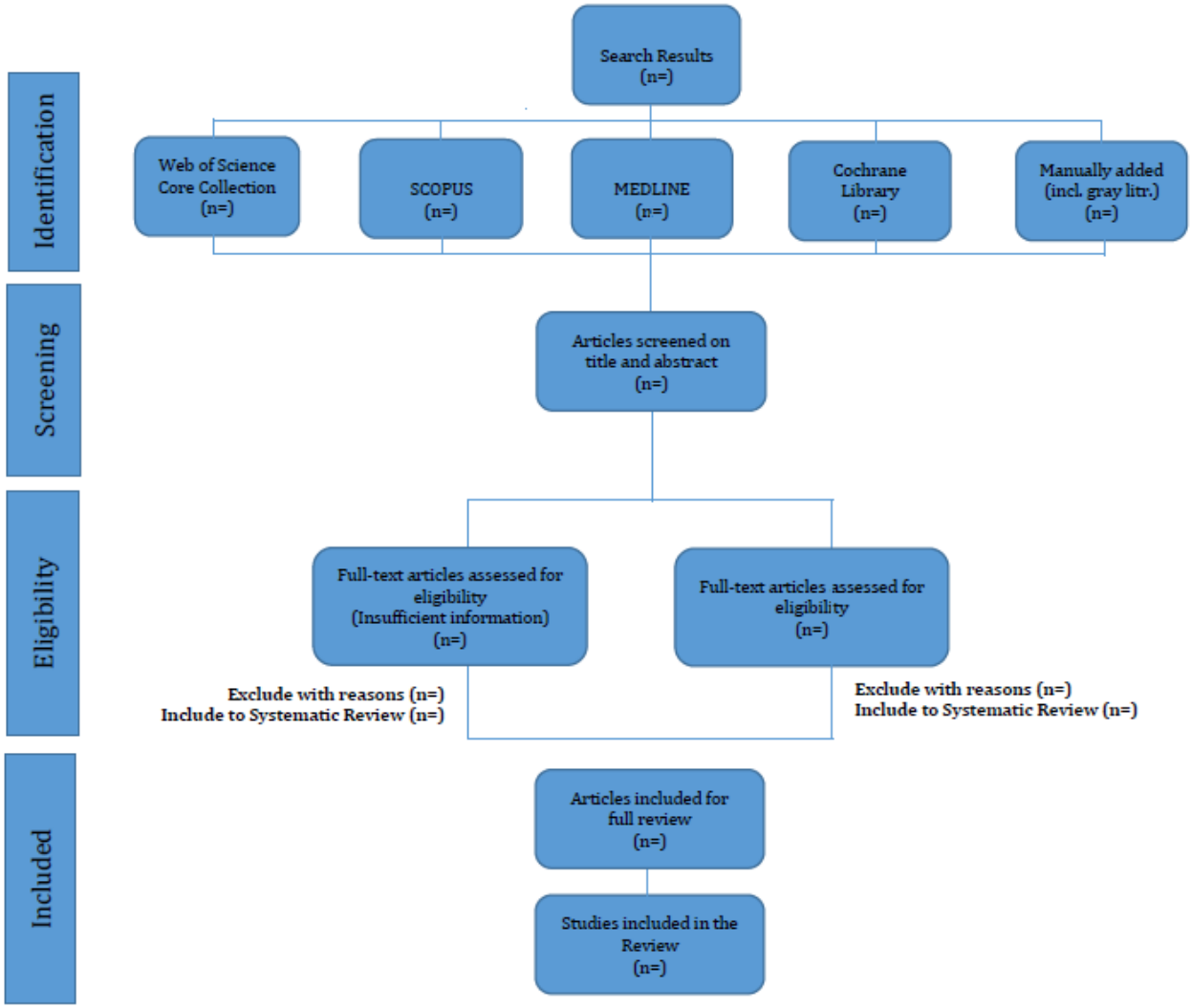
Data Sources and Search Methods

MEDLINE, Cochrane Library, Web of Science, and Scopus international electronic databases were searched on July 12, 2015, using the following search terms/query: ("e-government" OR "government" OR "department" OR "organization" OR "agenc*" OR "hospital*" OR "clinic*") AND ("social media" OR "Facebook" OR "Twitter" OR "YouTube") AND ("health" OR "Healthcare"). These databases were chosen for this systematic review because they cover a wide range of disciplines, including public health, medicine, technology, business, and social sciences. The search was not restricted to language, but the article selection was restricted to publication date; only studies published between January 1, 2004, and July 12, 2015, were included in this review. The year 2004 has been chosen as a starting point, since this was when Facebook, the most widely used social media website, was created. Relevant gray literature sources such as World Health Organization reports and working papers (searched via World Health Organization's Institutional Repository for Information Sharing), and industry (eg, consultancy and marketing) reports (searched via Google search engine) were also searched to identify relevant publications.

Studies Screening and Selection

One reviewer (AT) has independently screened generated citations with the help of EPPI-Reviewer 4 systematic review software (EPPI-Reviewer), and will extract the data from the qualified articles. Another author (CP) will then check this work to ensure accuracy. In case of any disagreement between reviewers during this stage, the arbitration of the third reviewer (MF) will be sought to resolve it. A PRISMA [37] flow diagram (Figure 3) will be created to track the study selection process and ensure transparency and replicability of the review. The final PRISMA diagram will specify the missing "n=" values, which will be the outcome of our analysis.

Figure 3. PRISMA flow diagram illustrating literature search and articles selection process.



Eligible Participants

Public sector health organizations that have deployed social media as a part of their e-government strategy including, but not limited health agencies, governmental departments and ministries. Public (state-funded) hospitals (when defined as such by publications' authors) will be also included in this review, to see whether there are any similarities or differences in how they use social media compared with other public health organizations.

Eligible Study Designs

This is not a review of clinical trials and our inclusion criteria encompass all types of study designs. However, based on our preliminary scoping work, we expect that most studies will be exploratory in nature. The inclusion and exclusion criteria are presented in Textbox 2.

Textbox 2. Inclusion and exclusion criteria.

Inclusion criteria

- Academic or industry research with a primary focus on the adoption and use of social media by public sector health organizations at the regional or national levels. For example, studies that focused on social media adoption by government departments of public health, regional health authorities, state-funded (public) hospitals or other government-sponsored agencies with a public health remit.
- Studies focused on the most popular social media sites, such as Facebook, Twitter, and YouTube [38-40].
- Studies published between January 1, 2004, and July 12, 2015.

Exclusion criteria

- Studies focused on private sector health organizations.
- Studies focused on individual units within public sector health organizations, such as emergency care or cardiology services and individual clinics.
- Studies primarily focused on social media for health surveillance or research.
- Studies focused on uses by specific professional or patient groups (eg, diabetes specialists or patients) or by individuals.
- Studies published before January 1, 2004, and after July 12, 2015.

Outcome Measures

Primary Outcomes

Indicators of uptake and use of social media and reported impacts on organizational transparency, efficiency, or effectiveness.

Secondary Outcomes

Perceived increase of government-to-government, government-to-citizen, government-to-business, and government-to-employee interaction, engagement, and satisfaction.

Data Analysis and Synthesis

We plan to narratively summarize and synthesize review results, taking into consideration the likelihood of heterogeneity of organizations and social media types studied, as well the designs of those studies. The following information is planned to be extracted from the studies that meet the inclusion criteria:

- authors/year;
- setting: country; organization; size; year;
- social media used;
- stated objective/purpose for using social media;
- research question;
- theoretical basis;
- study design and scope;
- outcomes examined, if relevant;
- main findings;
- conclusion/comments.

Critical Appraisal Techniques

Critical Appraisal Skills Programme checklist [41] will be used to assess the quality of the included studies.

Results

A comprehensive search strategy was created, tested, and applied to the aforementioned databases. Generated citations have been uploaded to EPPI-Reviewer, where they are being screened and coded. The work on this review is planned to be completed in the beginning of 2016 and its results will be presented at international academic conferences and published in a respected peer-reviewed journal.

Discussion

Social media present potentially valuable opportunities for public sector health organizations to meet the objectives of e-government, and social media are increasingly being used by such organizations. However, little is known about how this is being achieved in practice and no previous systematic reviews have sought to synthesize the relevant evidence. Undertaking a systematic review in this area is complicated by variability in the terms used to describe the concepts of e-government and public health, although taxonomies are available to support interpretation of the literature. The priorities for using social media in the health sector appear to be somewhat different from those of the generic e-government agenda, although the terms and concepts referred to in the relevant research literature overlap significantly. The preliminary observation that most research articles within the scope of our systematic review focus on usage alone suggests that public health organizations may be taking it for granted that social media will deliver benefits, rather than attempting to track their objectives for adoption or evaluate their impacts. A more detailed analysis of articles meeting the inclusion criteria will help to improve our understanding of this literature and inform the development of recommendations for research and practice. Undertaking this scoping review has provided valuable insights to guide the design and interpretation of the research literature with reference to a wider range of conceptual, disciplinary, and international considerations, supporting previous recommendations for the planning of systematic reviews [42-44] as well as testing innovative methods of mapping relevant terms and concepts.

Authors' Contributions

AT developed the search strategy and undertook the scoping exercises, with input from CP and MF. AT and CP will conduct the screening and apply the inclusion/exclusion criteria, with third-party arbitration by MF, where necessary. Included articles will be subjected to data extraction and synthesis by AT, with verification by the entire team. AT and CP co-wrote the manuscript, with input from MF.

Conflicts of Interest

None declared.

Multimedia Appendix 1

Existing systematic reviews on social media in health care.

[PDF File (Adobe PDF File), 393KB - resprot_v5i1e42_app1.pdf]

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Original Paper

A Systematic Review Protocol to Assess the Effects of Physical Activity on Health and Quality of Life Outcomes in Adolescent Cancer Survivors

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Abstract

Background: The benefits of physical activity for child and adult cancer survivors have been summarized in previous systematic reviews. However, no review has summarized the evidence for adolescent cancer survivors.

Objective: This paper describes the design of a protocol to conduct a systematic review of published studies examining the effects of physical activity on health and quality of life outcomes for adolescent cancer survivors.

Methods: Several guidelines informed the development of this protocol. The Preferred Reporting Items for Systematic Review and Meta-Analysis Protocols guidelines provided the structure by which to conduct and report the protocol; though some adaptations were made with regards to search terms, data synthesis, and evaluating the risk of bias. The Cochrane Handbook for Systematic Reviews of Interventions was used to guide research question development, search term selection, and the data extraction form. The Consolidated Standards of Reporting Trials guidelines helped inform the data extraction form. Lastly, the Guidance on the Conduct of Narrative Synthesis in Systematic Reviews informed the data synthesis. Ten electronic databases were identified and a search strategy was developed using a combination of Medical Subject Headings terms and keywords that were developed by the authors and peer reviewed by a university librarian. Both authors independently screened eligible studies for final inclusion, and data were abstracted using a form developed by the research team. A decision was made to synthesize all data narratively.

Results: The review has now been completed, peer-reviewed, and accepted for publication in a forthcoming issue of JMIR Cancer.

Conclusions: As this will be the first systematic review on this topic, outlining the protocol ensures transparency for the completed review. Further, this protocol illustrates how elements from several guidelines were incorporated to answer the research question (ie, what is the effect of physical activity on health and quality of life outcomes in adolescent cancer survivors). This flexible approach was necessary as a function of the paucity of available research on this topic.

(JMIR Res Protoc 2016;5(1):e54) doi:10.2196/resprot.5383

KEYWORDS

controlled clinical trial; randomized controlled trial; adolescent; oncology; neoplasm; exercise; quality of life.

Introduction

Over 7500 adolescents (defined herein as individuals between 13 to 19 years of age) living in North America are diagnosed with cancer each year [1,2] and become a cancer survivor. The National Cancer Institute defines a cancer survivor from the point of diagnosis onward [3]. As such, a cancer survivor may be actively receiving treatment (ie, on-treatment) or have completed treatment (ie, off-treatment). This definition of a cancer survivor will be used throughout this paper.

Approximately 80% will survive but will be at an increased risk for disability, morbidity, and mortality [4-8]; impaired physical, psychological, and social functioning [9,10]; and reduced quality of life [11,12]. In addition, normative growth, maturation, and development may be disrupted because of the disease and its treatments [13]. The National Comprehensive Cancer Network and the Institute of Medicine have identified adolescents and young adults as a distinct group of survivors who need to come to the forefront of efforts to lessen the impact of cancer [14,15]. Therefore, many researchers are taking action to identify complimentary therapies that can reduce the side effects of cancer and conventional cancer treatments.

Physical activity has been suggested as an effective adjunctive therapy to minimize the disruptions caused by cancer and its treatments [16]. Several reviews are available reporting on the safety and effects of physical activity on physical, psychological/emotional, and social health for child and adult cancer survivors [17-22]. However, no review has focused exclusively on adolescent cancer survivors despite acknowledgment that they are in a distinct developmental stage during their cancer experience [23]. In an effort to fill this gap, a review summarizing this information is needed. Thus, a systematic review protocol was developed. Both authors adhered to this protocol to comprehensively review and synthesize published studies focusing on the effectiveness of physical activity for promoting health and quality of life among adolescent cancer survivors. In doing so, adaptations from existing guidelines for systematic review protocols and systematic reviews (ie, Preferred Reporting Items for Systematic Review and Meta-Analysis Protocols, Cochrane Handbook for Systematic Reviews of Interventions, Consolidated Standards of Reporting Trials, Guidance on the Conduct of Narrative Synthesis in Systematic Reviews) were necessary because of the challenges of developing and conducting a systematic review on a topic with a paucity of available research. Therefore, the aim of this systematic review protocol was to outline the step-by-step process underlying the design and conduct of a systematic review exploring the effectiveness of physical activity for promoting health and quality of life in adolescent cancer survivors.

Methods

This review protocol followed the Preferred Reporting Items for Systematic Review and Meta-Analysis Protocols guidelines [24]. The guidelines consist of 17 items to facilitate the preparation and reporting of systematic review and meta-analysis protocols. Items cover 3 aspects of reporting: (1) administrative information (5 items), (2) introduction (2 items), and (3) methods (10 items). Specifically, adhering to Preferred Reporting Items for Systematic Review and Meta-Analysis Protocols guidelines from study inception promoted systematically defined concepts and inclusion criteria, provided a decision analytic framework, and offered guidance for writing up the final protocol. In addition, elements from the Cochrane Handbook for Systematic Reviews of Interventions, the Consolidated Standards of Reporting Trials, and the Guidance on the Conduct of Narrative Synthesis in Systematic Reviews were used as applicable. Each of these guidelines and the necessary changes (and the rationale for such changes) are provided below.

Inclusion Criteria

Studies were included if they were published in English in peer-reviewed scientific journals. The types of participants, interventions, outcomes, and studies that were considered for inclusion are described below.

Types of Participants

Studies were included if participants were adolescents aged 13 to 19 years who were diagnosed with any type of cancer and were at any point on the cancer trajectory (ie, diagnosis, treatment, post-treatment, palliation). Careful consideration was given in selecting this age range. Although there are several definitions currently being used, the ultimate goal of this review was to determine the effectiveness of physical activity for cancer survivors in their teenage years (ie, 13 to 19 years). Also, the definition selected adheres with recent efforts by other research groups [10]. Further, the decision to include cancer survivors on- and off-treatment is consistent with the National Cancer Institute's definition of a cancer survivor [3]. In cases where a wider age range was used, 50% or more of the sample had to contain participants meeting the above age criteria. This decision was made to increase the number of studies eligible for inclusion.

Types of Interventions

Physical activity was defined as consisting of aerobic training, resistance training, flexibility training, combined training, or any other form of physical movement with the goal of increasing energy expenditure. Physical activity interventions were limited to those that included more than one session. No restrictions were placed on where the intervention was delivered (eg, hospital, home, school, community), the format of the intervention (eg, group-based, individual), or the individual delivering it (eg, physiotherapist(s), exercise professional(s), researcher(s), personal trainer(s), parent(s)).

Types of Outcome Measures

Studies that included measures of health and/or quality of life as primary or secondary endpoints were included. Health outcomes included any participant-reported or objective assessment of physiological functioning (eg, fatigue, body composition, cardiovascular capacity, strength, flexibility). Quality of life outcomes included any participant-reported assessment of functioning across physical, psychological/emotional, and social domains.

Types of Studies

Only experimental study designs were included. Randomized controlled trials or controlled clinical trials were selected, as these study designs constitute the most robust form of clinical evidence [25]. Further, there had to be at least pre-post assessments. This decision was made so that any change observed over the course of the intervention could be more confidently attributed to the physical activity intervention [26].

Exclusion Criteria

Interventions that had multiple program features (eg, multiple behavior change strategies, nutrition counseling) were excluded, as any observed effects as a result of the intervention could not be attributed solely to physical activity. Furthermore, those with insufficient details on the target population, intervention, comparison condition, or outcomes (after study authors were contacted and it was determined the requested information was unavailable) were ineligible.

Data Sources and Search Strategy

A search strategy was developed using an iterative process based on recommendations from a university librarian (YL) and the methods sections (ie, keywords, procedures) from existing reviews on physical activity and cancer [18,20]. The strategy included a combination of Medical Subject Headings terms and keywords related to the population (eg, adolescent, young adult, young person, teenager, cancer, neoplasm), intervention (eg, exercise, physical fitness, aerobic exercise, resistance training, flexibility), comparison condition (eg, control groups, usual care), and outcomes (eg, health-related fitness, range of motion, quality of life, mood). During a preliminary search of MEDLINE, a limited number of studies were identified. Therefore, the search strategy was revised by excluding terms related to outcomes in order to reduce the likelihood of limiting the search to predefined outcomes and to maximize the number of studies retrieved (see Appendix 1 for the final MEDLINE search strategy). The revised search strategy was then translated and the following 10 electronic databases were searched from inception to November 2015: CINAHL, Cochrane Central Register of Controlled Trials, Embase, LILACS, MEDLINE, PEDro, Physical Education Index, PsycINFO, PubMed, and SPORTDiscus. Similar to other systematic reviews [17-21], following this, the reference lists of all studies meeting the inclusion criteria and any relevant reviews identified during the electronic database search were scanned to identify additional studies.

Study Selection

All studies identified in the database search were exported to a reference managing software [27] and duplicate records were deleted. Both authors independently reviewed the titles and abstracts of all references. Articles clearly not meeting the established inclusion/exclusion criteria were excluded. Following this, both authors independently screened the full text articles of abstracts identified to select the studies to be included. Then the reference lists of included studies and relevant reviews were scanned to identify additional studies. Both authors independently screened the full texts of these additional articles to determine inclusion/exclusion. Third-party arbitration (AJ and CO) was available to resolve any inconsistencies in the selection of studies for inclusion/exclusion. A Preferred Reporting Items for Systematic Review and Meta-Analysis flow diagram [28,29] was prepared to show the overall process of study selection and the number of citations reviewed at each stage of this review.

Data Collection

A data extraction tool was developed specifically for this review based on recommendations provided in the Cochrane Handbook of Systematic Reviews of Interventions [30]. In cases where details were missing on study design, population, intervention, or outcomes, the authors of included studies were contacted by email. After the first contact attempt, if no response was received, the study authors were contacted 2 more times approximately 3 to 4 weeks apart. The following information was extracted from each included article: (1) sources of data, (2) study design and study period, (3) characteristics of the population (ie, number of participants randomized, age, type(s) of cancer diagnosed, cancer phase), (4) intervention characteristics (ie, supervision, setting, length, frequency, intensity, activity type(s)), (5) outcome measures (ie, health and/or quality of life), and (6) outcomes (ie, health and/or quality of life).

In addition to extracting the above standard data, additional information was documented on the use of theoretical frameworks (ie, whether the study was informed by theory). The use of intention-to-treat analysis was also recorded since intention-to-treat analyses generally provide an unbiased estimate of treatment effect. Intention-to-treat is a well-regarded approach to the design, conduct, and analysis of a trial [31], and it is a key component in the Consolidated Standards of Reporting Trials guidelines [32]. Additionally, data on variables not considered to be health and/or quality of life outcomes (eg, intervention acceptance, adverse events, adherence to the study protocol) were extracted to provide a more comprehensive understanding of the state of the literature.

Data Synthesis

Given that the main purpose of the systematic review was to comprehensively review and synthesize published studies focusing on the effectiveness of physical activity for promoting health and quality of life among adolescent cancer survivors, all data extracted from the articles were presented narratively in text and summary tables. This decision was made because narrative synthesis provides a broad overview of relevant information through a textual approach and is appropriate when it is expected that studies will be too heterogeneous to allow for a quantitative summary [33]. Heterogeneity of studies was assessed according to content, rather than by performing statistical tests for homogeneity. It was expected that the studies included in this review would vary widely. This was based on a recent review conducted with pediatric cancer survivors that found large variability across studies [17]. To ensure the quality of the narrative synthesis, the Guidance on the Conduct of Narrative Synthesis in Systematic Reviews was followed as appropriate to accurately report the review search results and analysis summary [33]. Specifically, the included studies were carefully reviewed and the limitations of each (ie, quality assessment) were described. Additionally, the entire data extraction and synthesis process was carefully detailed, and objective third-party review (AJ) was utilized.

Results

The review has now been completed, peer-reviewed, and accepted for publication in a forthcoming issue of JMIR Cancer [34].

Discussion

Whereas other recent systematic reviews were undertaken to investigate the benefits of physical activity for children and adults diagnosed with cancer [17-20], none have focused exclusively on adolescents. Further, few, if any, have published a detailed protocol either independently or as supplemental material. Thus, this protocol adds to the field of physical activity and cancer. A key strength of this review protocol is the use of multiple gold standard guidelines. By incorporating different elements from each guideline, a solid framework and structure was created by which the research question could be answered. Additional strengths are the inclusion of a university librarian (YL) with experience conducting systematic reviews who assisted with peer-reviewing the search strategy, the application of a data extraction template, and a flexible approach to data acquisition and synthesis.

Notwithstanding the strengths, there were key challenges to preparing and finalizing this review protocol. First, there were challenges formulating the inclusion/exclusion criteria. In general, given the complexity and breadth of definitions for adolescent cancer survivors and the numerous iterations of physical activity interventions, careful consideration was given to ensuring the best evidence was identified to answer the research question. Second, identifying pertinent literature was difficult given the paucity of results obtained in preliminary tests of the search strategy. A flexible approach to search terms and keywords was necessary to ensure more studies were identified for review. Third, in light of the lack of research focused on adolescent cancer survivors, synthesizing and interpreting data was assumed to be a challenge. Thus, a narrative approach to data synthesis was selected. New topics pose inherent challenges for systematic review protocol development. As the literature in this area grows, so will the opportunities to refine protocols, re-run searches, and update findings. However, until then, protocols should be disseminated to ensure transparency of completed reviews and aid other researchers in developing their review protocols.

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AW and JB conceptualized and designed the review protocol, performed the systematic literature searches, screened and selected studies, and extracted and interpreted the data. They were involved in all aspects of drafting, revising, and finalizing this protocol manuscript. Further, both approved the order of authorship.

Conflicts of Interest

None declared.

Multimedia Appendix 1

MEDLINE search strategy.

[PDF File (Adobe PDF File), 33KB - resprot_v5i1e54_app1.pdf]

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Original Paper

Online Tobacco Cessation Training and Competency Assessment for Complementary and Alternative Medicine (CAM) Practitioners: Protocol for the CAM Reach Web Study

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Abstract

Background: Complementary and alternative medicine (CAM) practitioners, such as chiropractors, acupuncturists, and massage therapists, are a growing presence in the US health care landscape and already provide health and wellness care to significant numbers of patients who use tobacco. For decades, conventional biomedical practitioners have received training to provide evidence-based tobacco cessation brief interventions (BIs) and referrals to cessation services as part of routine clinical care, whereas CAM practitioners have been largely overlooked for BI training. Web-based training has clear potential to meet large-scale training dissemination needs. However, despite the exploding use of Web-based training for health professionals, Web-based evaluation of clinical skills competency remains underdeveloped.

Objective: In pursuit of a long-term goal of helping CAM practitioners integrate evidence-based practices from US Public Health Service Tobacco Dependence Treatment Guideline into routine clinical care, this pilot protocol aims to develop and test a Web-based tobacco cessation training program tailored for CAM practitioners.

Methods: In preparation for a larger trial to examine the effect of training on CAM practitioner clinical practice behaviors around tobacco cessation, this developmental study will (1) adapt an existing in-person tobacco cessation BI training program that is specifically tailored for CAM therapists for delivery via the Internet; (2) develop a novel, Web-based tool to assess CAM practitioner competence in tobacco cessation BI skills, and conduct a pilot validation study comparing the competency assessment tool to live video role plays with a standardized patient; (3) pilot test the Web-based training with 120 CAM practitioners (40 acupuncturists, 40 chiropractors, 40 massage therapists) for usability, accessibility, acceptability, and effects on practitioner knowledge, self-efficacy, and competency with tobacco cessation; and (4) conduct qualitative and quantitative formative research on factors influencing practitioner tobacco cessation clinical behaviors (eg, practice environment, peer social influence, and insurance reimbursement).

Results: Web-training and competency assessment tool development and study enrollment and training activities are complete (N=203 practitioners enrolled). Training completion rates were lower than expected (36.9%, 75/203), necessitating over enrollment to ensure a sufficient number of training completers. Follow-up data collection is in progress. Data analysis will begin immediately after data collection is complete.

Conclusions: To realize CAM practitioners' potential to promote tobacco cessation and use of evidence-based treatments, there is a need to know more about the facilitative and inhibitory factors influencing CAM practitioner tobacco intervention behaviors (eg, social influence and insurance reimbursement). Given marked differences between conventional and CAM practitioners, extant knowledge about factors influencing conventional practitioner adoption of tobacco cessation behaviors cannot be confidently

extrapolated to CAM practitioners. The potential impact of this study is to expand tobacco cessation and health promotion infrastructure in a new group of health practitioners who can help combat the continuing epidemic of tobacco use.

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KEYWORDS

tobacco cessation; brief intervention; online training; communication; acupuncture; chiropractic; massage therapy

Introduction

Complementary and alternative medicine (CAM) practitioners such as chiropractors, acupuncturists, and massage therapists are a growing presence in the US health care landscape as a significant proportion of Americans report using CAM. The National Health Interview Survey (NHIS) periodically includes additional questions (Adult Alternative Medicine (ALT) supplement) on the use of CAM therapies. Clarke et al [1] examined trends in CAM use from the NHIS 2002, 2007, and 2012 ALT supplements, adjusting for methodological differences across the three surveys. They reported that the percentage of adults who had used any form of CAM in the preceding 12 months ranged from 32.3% in 2002 to 35.5% in 2007, and 33.2% in 2012. Among populations known to be at greater risk for tobacco use (eg, poor, lower educational attainment, public health insurance, and uninsured) there were a significant percentage using CAM in 2012. They found that 20.6% of poor adults, 24.4% of adults with a high school diploma or GED, 24.8% of public insurance, and 22.9% of uninsured adults reported using CAM [1]. An analysis of 2007 NHIS data found that significant numbers of respondents who used CAM services in the prior year had the following chronic disease and/or major risk factors: (1) current smoking (17.4%), (2) hypertension (18.1%), (3) obesity (21.4%), and (4) physical inactivity (22%) [2].

Tobacco use accounts for nearly 443,000 deaths in the United States each year and is responsible for US $96 billion in health care expenditures annually [3], yet use of effective cessation aids remains low. Of the 45.3 million tobacco users in the United States in 2010 [4], 52.4% reported a quit attempt in the previous year [5,6]. Overall, nearly 69% of smokers report they want to quit [7]. Effective tobacco cessation treatments recommended by the US Public Health Service Guideline on Treatment of Tobacco Dependence (PHS Guideline) [8], are more widely available than ever, and yet are still greatly underutilized. Nearly 70% of tobacco users attempt to quit without assistance [7] and unaided attempts are rarely successful [9]. There is an urgent need to increase use of proven cessation aids.

Decades of public health tobacco control efforts have led to steady declines in tobacco use prevalence from 40.3% in 1964 to 19.3% in 2011, and 19.2% in 2013 [4,10,11]. But in recent years inconsistency or stagnation in this downward trend has prompted public health calls for new and expanded strategies for approaching tobacco cessation, including broadened insurance coverage for cessation treatment [5]. CAM practitioners have the opportunity to intervene with chronic disease risk factors such as tobacco use, and in at least one study, chiropractors were found to be more likely to engage in tobacco cessation activities with patients than primary care physicians [12].

With a few exceptions [13-16], CAM practitioners have been largely overlooked in the nation's tobacco control agenda. For more than two decades, public health efforts have targeted physicians for tobacco brief intervention (BI) training [17]. Only more recently has BI training been offered to other biomedical health care professionals (nurses, dentists, pharmacists) [17]. Despite clear evidence that asking, assessing, and intervening by health care providers results in increased quit rates [8], biomedical physicians do not consistently offer tobacco BIs. An Association of American Medical Colleges survey found that 86% of physicians advise patients to stop tobacco use but only 37% discuss counseling options, 31% recommend nicotine replacement, and 13% refer patients to others for cessation treatment [18]. There is a clear need to expand research on tobacco cessation training beyond conventional health practitioners to increase the potential public health reach and impact of brief tobacco cessation interventions.

Web-based training has clear potential to meet large-scale training dissemination needs. Yet, despite widespread use of Web-based instruction for training health professionals [19], few online tools are available for assessing clinical skills competency**.** While initial development costs are higher for online training, costs for ongoing training delivery are low. Compared with classroom instruction, online training offers additional advantages including greater learner accessibility, increased convenience, and greater scalability. However, online evaluation tools for assessing clinical skills competency with similar accessibility, scalability, and low dissemination costs are still lacking. In preparation for a larger validation study, this study aims to create an online, scalable tool to assess clinical skills competency among CAM practitioners and to assess the feasibility of comparing this tool's performance to a live video standardized patient method of learner assessment.

Innovative aims of this study include (1) shift the current public health and clinical practice paradigm in tobacco cessation training, currently focused on conventional biomedical practitioners, to include training CAM practitioners in evidence-based knowledge and skills to promote tobacco cessation and encourage use of effective treatments; (2) develop a unique, Web-based tobacco cessation BI training program for CAM practitioners to improve their competency in helping their clients or patients quit tobacco; (3) develop a new Web-based tool to assess CAM practitioners' clinical skills competency at providing tobacco BIs; and (4) fill a gap in the literature by collecting new information on CAM providers regarding online BI training, clinical competency testing, and the facilitating and inhibiting role of practice environment, insurance reimbursement, and training availability on adoption of tobacco

cessation BI behaviors. Initial components of the study that have already been completed are described in the Results section. We note that the three CAM disciplines participating in the CAM Reach Web (CAMR-Web) study customarily use different terms to refer to persons seeking their care. Chiropractors and acupuncturists usually refer to "patients", whereas massage therapists usually say "clients". For simplicity, we will use "patients" throughout this paper.

Methods

The CAMR-Web study is a mixed-method (qualitative and quantitative) pilot feasibility study. Theoretical frameworks informing this work are Social Cognitive Theory [20], Adult Learning Theory [21,22], and Swing's [23] competency-based education and assessment framework, which informs the competency assessment tool development. It builds upon the BI curriculum and training development work completed in our prior CAM Reach (CAMR) study (NCI 1RO1 CA137375) [15,16]. The CAMR-Web study has three overlapping phases: Phase 1 (development of training website and competency assessment tool (completed)), Phase 2 (training website pilot study and assessment tool pilot validation study (underway), and Phase 3 (data synthesis and write-up (planned)).

CAM practitioners differ from conventional practitioners in substantive ways such as professional training, scope of practice, philosophies of practice and approaches to healing; practice patterns, and patient or client relationships [12,24-26]. Accordingly, the numerous tobacco cessation-training programs available to conventional practitioners are not well suited for CAM practitioners. In our previous CAMR study [15,16,27,28], we developed an in-person training workshop and practice system intervention that is uniquely and specifically tailored for CAM practitioners. It is based on extensive formative research with expert review and input from leaders in CAM education and research, tobacco cessation interventions and policy, and integrative medicine [16]. The CAMR study results showed that CAM practitioners had significant increases in tobacco cessation activities, motivation and confidence in helping patients quit tobacco, and comfort with providing information and referrals for PHS Guideline-based tobacco cessation aids. These increases occurred across all three practitioner types and were sustained at 12 months, despite heterogeneity in professional training, practice patterns and organization, and practice business models. Adapting the CAMR training for Web-based delivery was intended to create a unique tobacco BI training that is more accessible and scalable.

Despite growth of Web-based training of health care practitioners, effective Web-based methodologies to assess clinical competencies lag behind. "Standardized patient" is an evaluation method widely used in medical education for assessing clinical skills competency [29]. It most closely simulates a real patient encounter and is thus a major component of high-stakes exams (eg, Objective Structured Clinical Examination (OSCE) required for certification and licensure) [30,31]. Use of OSCEs and standardized patients in CAM practitioner education and evaluation is relatively new and uncommon; however, there is a gap in the literature on the use of these methodologies in CAM education and competency assessment. This study was designed to fill a need for innovative, scalable, Web-based tools to evaluate clinical skills competency that can approximate simulation of in-person methods such as a standardized patient.

Phase 1 (Complete)

Phase 1 of the study (completed) involved evaluation of the original CAMR study training curriculum for the adaptation necessary for Web-based training delivery of the content, followed by design and production of the training website. Specific methods for accomplishing these aspects are detailed in the following sections.

Design and Production of Training Website

The curriculum and Web development teams worked collaboratively in the creation of the CAMR-Web training content map and specifications to best suit the curriculum. This team of investigators and staff had extensive experience working together to develop online tobacco cessation trainings. The content map and specifications served as a guide for the design process which began with storyboarding interactive learning activities and course content, followed by developing "screenplay" versions of learning activities and content and identifying needed animation or video assets and/or interactive simulations to reinforce and demonstrate desired instructional objectives within the content map. Database connectivity was designed to facilitate (1) the collection of demographic data from users in a convenient, easy-to-analyze format; (2) the use of open- and closed-ended evaluation measures; (3) the collection and instantaneous scoring of nominal and ordinal evaluation data; (4) the ability to track user progress through curriculum modules and the site as a whole; and (5) the ability to deploy dynamic, customized Web pages created in real-time for each user.

Adaptation of CAMR Training Content for Online Delivery

Investigators adapted the existing CAMR in-person, multimedia training to best suit the online learning environment. Continuing the highly successful, inclusive approach to developing the CAMR in-person curriculum, adaptation of the curriculum for Web-based delivery was conducted in consultation with external reviewers from our local and national advisory panels. Upon project startup, local advisors were invited from our pool of completed CAMR study participants. In tandem with this curriculum content conversion, we developed customized clinical cases to assess core competencies addressed by the CAMR Web online curriculum. We then deployed the clinical cases through the DecisionSim simulation development learning platform [32], integrating the clinical cases in such a way as to provide for incremental and summative learner competency assessment as described below.

Adaptation of CAMR Instruments and Measures for Online Data Collection

Evaluation instruments including screening, baseline, pretraining and post-training assessments, and follow-up surveys were developed or adapted from existing instruments developed for

the CAMR study [16,27,33]. They were pilot tested with individuals from the target audiences and reviewed by our advisory panels. User interface and database back-end components were constructed once instruments were finalized.

Development and Production of Simulation Cases and a Web-Based Competency Assessment Tool

Using DecisionSim, the summative assessment is an online simulation of a case-based practice of communication skills and application of tobacco cessation knowledge. The learner is presented with a tobacco-using patient or client scenario for which an "ideal" interaction path has been previously specified by investigators, in consultation with our advisors. Each response option is associated with a tag (in the back-end database) as being "optimal", "critical", or "poor." To successfully complete the training a learner needs to meet a minimum level of competence as defined by selection of a response path that is within a specified range of the "ideal" interaction path. To evaluate the online competency tool, we also developed a standardized patient "live simulation" role play exercise in which participants role play the exercise scenarios with a standardized patient via Skype video. Live simulation cases reflect a range of patients' readiness to quit and evaluate the practitioner's ability to assess the patient or client and adjust their BI behaviors accordingly. Standardized patient cases, scripts, and examiner checklists for the feasibility study were developed in tandem with simulation cases for the competency assessment tool. For both the live and online simulation cases, we developed related evaluation criteria and thresholds for determining competency. Development of evaluation criteria and thresholds were guided by published, evidence-based tobacco cessation behavioral support competencies and guidance documents (eg, those supported by national tobacco cessation programs [34]) and review and feedback from our advisory panels.

User, Usability, and Beta Testing of the CAMR Website and Competency Assessment Tool

The CAMR website underwent rigorous user and usability testing with local and national panel members as well as with members of the target population of CAM practitioners. Investigators reviewed the initial version of the training program and made modifications to improve usability. The initial version was also thoroughly tested in-house for code errors. It was tested for usability with volunteer CAM practitioners (N=18; 6 chiropractors, 3 massage therapists, 9 acupuncturist) with protocol analysis. Practitioners were given a set of tasks to mimic real-world use (eg, registering on the site, viewing the training, completing the competency assessment). Testers completed a postsurvey on ease of use and attractiveness of screen design [35]. Investigators and programmers reviewed the results and made changes to improve usability.

Near completion, the CAMR website was beta tested with CAM practitioners and advisory panel members (N=12). The goal of beta testing was to confirm that revisions made in response to usability study findings eliminated identified problems and that all site features function as intended. Beta testing also gauged the instructional effectiveness of the curriculum. The process followed protocols suggested by Trollip and Alessi [36], which included (1) beta testers have content expertise matching the expected, pre-existing knowledge base of the prototypical learner; (2) content quizzes employing the post-test instruments are used to conduct item analysis of online quizzes and learning activities and to gauge the effectiveness of the Web-based curriculum; and (3) testers are interviewed to identify attitudes toward the instructional experience.

Phase 2 (Underway)

Feasibility Study of CAMR-Web-Based Training

The feasibility study design is a single group with participant assessments at (1) baseline (study enrollment) and pretest prior to CAMR-Web training intervention; (2) immediately post-training; and (3) 3 and 6 months post training completion. A subsample of practitioners from each practitioner type was selected based on specific criteria to complete an in-depth, open-ended response follow-up survey on selected factors influencing practitioner adoption of tobacco intervention behaviors (eg, practice environment, peer social influence, and insurance reimbursement). Phase 2 participant enrollment is complete and data collection is still in progress.

Participant Recruitment, Eligibility, and Enrollment

Based on literature regarding attrition among online learners [37,38], and our own experience delivering online training to other target populations [39,40], we anticipated that roughly 50.0% (60/120) of participants would complete all parts of the proposed study activities. Given this estimate, we originally planned to recruit a total of 120 practitioners (40 per CAM discipline), with the goal of retaining at least 19 practitioners from each discipline through the end of the study. We recruited practitioners through websites, listservs, and newsletters of CAM practitioner professional organizations, alumni of schools of chiropractic, acupuncture, and massage therapy, and through the national provider network of a large third-party payer for chiropractic, acupuncture, and massage therapy services. Interested practitioners were directed to the project website for information about the study and access to screening and enrollment instruments.

To participate, practitioners must be ≥18 years of age, have unrestricted license or credentials to practice their CAM discipline, be actively in practice three quarters time to full-time, see at least 10 patients per week, have access to a computer with broadband Internet access and audio output, and be willing to provide consent and participate in the entire study. Practitioners were excluded if they reported participation in formal tobacco cessation training in the past 2 years.

Practitioners arriving at the project website were presented with a project overview. To enroll, practitioners first completed the screening instrument to determine eligibility. Responses to the screening items were processed immediately and the eligibility status displayed to the practitioner. Eligible participants were directed to a link to the consent portal. This portal displayed the consent document in its entirety with a link to download and print the form. To continue with enrollment, the participant was required to click a button indicating that he or she has read and agrees to all parts of the online consent form. Next, the online baseline and pretraining survey were presented for

completion. (Textbox 1). Once completed, participants were enrolled and provided full access to the CAMR-Web training.

Participant Training (Completed) and Follow-Up (in Progress)

Enrolled practitioners experience the CAMR-Web tobacco cessation BI curriculum [16], which was tailored for CAM practitioners and focuses on a nonconfrontational approach that is referred to as a "helping conversation." In our experience developing the in-person CAMR curriculum [16], transforming the "5 A's" protocol [8] into a less proscriptive and more motivational framework is more acceptable to these CAM practitioner groups. Practitioners are encouraged to complete the CAMR-Web training within 4 weeks. Upon finishing the training, practitioners are asked to complete the online competency assessment tool and post-training assessments. A training certificate is issued to all practitioners who successfully demonstrate the requisite competency level and who respond correctly to at least 80% of knowledge items. Practitioners receive compensation for completing the research assessment and eight continuing education credits for completing the online training. Practitioners also receive a training completion packet by mail with twenty copies of each patient handout. They may request additional copies as needed throughout their study participation.

After training, practitioners are asked to complete Web-based questionnaires at 3 and 6 months post training completion (Textbox 1). These follow-up intervals are based on our prior work with community-based tobacco cessation training of nonpractitioners and on the current CAMR study. Follow-up in less than 3 months does not allow sufficient time for participants to have implemented their training, while changes in self-efficacy measures tend to stabilize by 6 months. Participants receive up to US $110 incentive for completion of all study assessments.

Textbox 1. Summary of CAMR evaluation instruments, measures, and domains.

Measure and addressed domains

- Screening
  - Eligibility criteria
- Baseline and pretest assessment
  - Demographics
  - Cessation intervention behaviors, attitudes regarding tobacco use, quitting, and intervening with patients
  - Confidence and self-efficacy in performing intervention behaviors
  - Training-related knowledge [27]
- Post-training assessment
  - Cessation intervention behaviors, attitudes regarding tobacco use, quitting and intervening with patients,
  - Confidence and self-efficacy in performing intervention behaviors,
  - Training-related knowledge,
  - Training site usability [35].
- Web-based skills competency assessment
  - Ability to recognize and select appropriate: (1) Techniques to ask or become aware of patient or client tobacco use; (2) Communication skills to assess tobacco use of client/patient; (3) Advice and encouragement to consider quitting tobacco; (4) Assistance (eg, referral to guideline based services and provision of materials or information); (5) Arrangements to follow up with patient or client regarding tobacco use.
- Follow-up assessments at 3 and 6 months
  - Cessation intervention behavior and use of training materials,
  - Confidence and self-efficacy in performing intervention behaviors
- In-depth, open-ended follow-up survey
- Open-ended, in-depth survey with a subsample of participants

Competency Assessment Tool Validation Feasibility Pilot

This feasibility pilot study is using the University of Arizona College of Medicine's standard protocols for training standardized patients. To test feasibility for a larger validation study, we recruited a subsample of 10 CAM practitioners who completed the CAMR-Web training (n=3-5 per practitioner type) to participate in the live video standardized patient interviews to assess CAM practitioner skill competency. Eligible

participants must have completed training within the prior 2 weeks and have access to bi-directional Internet video capability (eg, Skype) sufficient to participate in a 30-minute, live video, standardized patient role-play session. Each participant is evaluated with two standardized patient cases (1 ready to quit, 1 not ready to quit), and each case is scored separately. Practitioners are not offered feedback on their performance to minimize the impact of the standardized patient on practitioner behavior during the remainder of the follow-up period. Practitioners who complete the role-play are then invited to complete a survey about usefulness of the activity. Their live assessment scores are then compared to their online competency assessment scores.

In-Depth, Open-Ended Response Survey of Practitioner Subsample

Approximately 6 months after training completion, practitioners are screened for eligibility to complete an in-depth online survey with open-ended response options. Inclusion criteria into this survey require that (during their 3-month follow-up survey) practitioners report seeing tobacco users in their practice and respond with "always" or "often" on a 4-point scale (always, often, sometimes, never) to at least 3 of the following (1) assessing interest in quitting; (2) identifying reasons to quit; (3) offering materials; (4) discussing medications; (5) treating tobacco addiction; or (6) referring to tobacco treatment outside of their practice.

Open-ended responses from the online survey will be analyzed using qualitative methodology to gain a more detailed understanding of practitioners' experiences as they apply training in real-world settings [41]. Open-ended survey questions were designed to gather formative data on specific factors that might influence CAM practitioner adoption of tobacco BI behaviors to inform a future dissemination trial. Investigators used interview guides from the CAMR study to design items focused on practice environment, peer social influence, and third-party reimbursement. Question themes focused on (1) facilitating or inhibiting factors in practice environment; (2) desirability and utility of peer social networking; and (3) reimbursement issues such as eligibility, qualifications and credentialing, documentation, and price-point. Reimbursement themes will be explored only at the end of the study to minimize potential effects on practitioner behavior during the post-training observation period.

Sample Size

Data for power analysis came from pretest and post-test assessments of knowledge and confidence (self-efficacy) in the previous in-person CAMR training study. We compared the number of correct answers on a 15-item tobacco cessation knowledge test administered before and after training for 33 practitioners. Scores on the items approximated a normal distribution. A paired *t* test analysis indicated significant improvement in knowledge (t=6.7, P<.001). Using the data from the 33 practitioners (mean (SD) of the paired differences is 2.1(1.8)), a *t* test power analysis indicated that 10 subjects would be needed for power =.90 and alpha =.05 and 11 subjects needed for the Wilcoxon test which makes fewer assumptions about the distribution of the scores than does the *t* test and typically requires a larger sample.

The pretest and post-test confidence question was "I am confident that I can personalize the benefits of quitting with each individual patient". The possible responses ranged from 1 ("Not at all confident") to 4 ("Very confident"). The scores on this question approximated a normal distribution. A paired *t* test analysis indicated a significant positive change (t=5.9, P<.001). Using the data from the 33 practitioners (mean (SD) of the paired differences is 0.7(0.84)), a *t* test power analysis indicated that 18 subjects would be needed for power = .90 and alpha = .05. For the Wilcoxon test, 19 subjects would be needed. Power analyses were conducted using PASS (Version 11.0.8) from NCSS Statistical Software.

Phase 3 (Planned)

The process of using the CAMR-Web training website is being evaluated by assigning CAM providers IDs, which mark usage in the backend database, including time spent, pages viewed, and modules completed. The association of use of the website with change in knowledge and confidence will be assessed using regression analysis methods. These analyses will be limited by use not being randomly assigned so potential third variables (eg, demographics, years in practice, patient volume) will be included as covariates.

In so far as knowledge and confidence were measured at baseline, 3, and 6 months (for three practitioner types), changes will be analyzed using mixed-models [42]. Adjustments will be made for participant characteristics (eg, demographics, years in practice, patient volume, and provider group for each CAM group). This approach will enable us to simultaneously assess intervention effects for participants overall and by group. The analysis will be conducted using SAS 9.4 [43]. Intra-class correlation coefficients will be calculated between CAM providers' knowledge and confidence measures, and clinical skills scores after completing the new DecisionSim Web-based CAMR competency tool and the live standardized patient assessment. Positive correlations at P<.05 were planned to establish predictive validity.

Qualitative analysis of open-ended survey responses will use a coding-categorizing technique [44,45]. This qualitative strategy is a form of content analysis that involves arranging the data into categories sorted by broader themes (eg, patient presenting symptom, events, etc) to assess issues not easily captured by closed survey questions and to generate themes for further study.

Results

We have enrolled 203 practitioners (63 chiropractors, 78 acupuncturists and traditional Chinese medicine practitioners, and 62 massage therapists). Initial training completion rates varied by CAM discipline, but overall the average completion rate was lower (36.9%, 75/203) than we expected based on our prior experience with other Web-based training. We therefore overenrolled to meet the original target of 20 practitioners per CAM group completing training and 3-month follow-up. At the time of writing, all training activities have been completed and follow-up survey data collection is in progress. Final data

collection will be completed in November 2015. In Phase 3, we will analyze the main outcomes, synthesize results from quantitative and qualitative data, evaluate the feasibility of a larger competency assessment tool validation study, and address the feasibility outcomes in preparation for a larger dissemination study.

Discussion

Principal Findings

Focusing tobacco BI training on conventional practitioners limits BIs' potential public health impact. Unhealthy lifestyle behaviors are the root cause of the growing burden of chronic disease in the United States [46-49]. Tobacco use, diet, and physical activity are three lifestyle behaviors affecting the nation's public health that are major risk factors for the most prevalent chronic diseases: cardiovascular disease, diabetes, and cancer. The Institute of Medicine report on CAM use in the United States found little research on the role of CAM in addressing national public health priorities requiring behavioral change [50]. This has also been recognized by the naturopathic physician and chiropractic communities [2]. Key questions remain regarding (1) the role of CAM practitioners in fostering and sustaining behavior change around tobacco use and other health behaviors; (2) CAM practitioners' behaviors related to promoting healthy behavior; (3) patients' use of CAM practitioners to support behavior change; (4) the potential role of CAM practitioners in preventive and promotive health; and finally (5) whether CAM practitioners can be engaged to fully participate in a public health community of practice [51]. Tobacco cessation can serve as a model with which to examine these issues.

Factors impacting CAM practitioner tobacco cessation behaviors are not well studied, especially regarding adoption of PHS Guideline-based tobacco dependence treatment practices. Among conventional practitioners, training [17], practice environment system changes [52-54], and pro-adoption peer social influences have generally been shown to increase tobacco BI behaviors, particularly in combination with one another [55]. Prior to this study, however, there was little to no research on how these factors affect CAM practitioners. Effects of insurance reimbursement on conventional practitioner tobacco BI behavior are mixed [56,57], and in the case of Medicare reimbursement evidence suggests underutilization of billing for cessation counseling services [58]. Insurance reimbursement for tobacco cessation services is now widely available for conventional medical practitioners, although not presently for CAM practitioners. Given this, and other substantive differences, extant knowledge about factors promoting conventional practitioners' adoption of tobacco intervention behaviors cannot be confidently extrapolated to CAM practitioners.

Conclusions

CAM practitioners are an increasingly important presence in the US health care system and already provide health and wellness care to significant numbers of patients who use tobacco and/or have other chronic disease(s) and risk factors. There has been scant research, however, into the potential factors impacting on CAM practitioners' willingness and ability to adopt evidence-based tobacco cessation behaviors as part of routine clinical practice. These factors include cessation training adapted to their professional discipline, professional scope of practice, and approaches to health and wellness. Other potential factors include practice organization and business models, and the availability of third party insurance reimbursement for tobacco cessation services. The keys to insurance reimbursement are proven competency and credentials, and evidence-based deployment of an effective intervention. To engage CAM practitioners in tobacco cessation, it is critical to know how best to increase incorporation of evidence-based interventions and practices into their routine clinical care. Similarly, research on the effect of insurance reimbursement is critical to develop evidence-based policy for CAM practitioners to provide effective cessation interventions. The potential impact of this study is to build infrastructure in a new group of health practitioners who can help combat the continuing epidemic of tobacco use.

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Authors' Contributions

All authors contributed to the study conceptualization and participated in its design and intervention development. MM, AH, and EE helped draft the manuscript. All authors read and edited for significant intellectual content and approved the final manuscript.

Conflicts of Interest

None declared.

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Abbreviations

ALT: Adult Alternative Medicine
BI: brief interventions
CAM: chiropractors, acupuncturists, and massage
CAMR: CAM Reach
CAMR-Web: CAM Reach Web
NHIS: National Health Interview Survey
OSCE: Objective Structured Clinical Examination
PHS: Public Health Service

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Protocol

Health Systems Readiness to Manage the Hypertension Epidemic in Primary Health Care Facilities in the Western Cape, South Africa: A Study Protocol

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Abstract

Background: Developing countries are undergoing a process of epidemiological transition from infectious to noncommunicable diseases, described by the United Nations Secretary General Ban Ki-Moon as ‘‘a public health emergency in slow motion.” One of the most prevalent in sub-Saharan Africa is hypertension, which is a complex chronic condition often referred to as a “silent killer” and key contributor to the development of cardiovascular and cerebrovascular diseases. Hypertensive patients in this setting are estimated to increase from 74.7 million in 2008 to 125.5 million in 2025, a 68% increase. However, there is an important gap between emerging high-level policies and recommendations, and the near-absence of practical guidance and experience delivering long-term medical care for noncommunicable diseases within resource-limited health systems.

Objective: To address this gap, our study will consist of field investigations to determine the minimum health systems requirements to ensure successful delivery of antihypertensive medications when scaling-up interventions to control the hypertension epidemic.

Methods: A cross-sectional analytic study will be conducted in the Western Cape using a mixed-method approach with two semistructured interview guides. The first will be for health professionals involved in the care of hypertensive patients within at least 6 community health centers (3 urban and 3 rural) to understand the challenges associated with their care. The second will be to map and assess the current supply chain management system of antihypertensive medications by interviewing key informants at different levels of the processes. Finally, modeling and simulation tools will be used to understand how to estimate minimum numbers of health workers required at each supply chain interval to ensure successful delivery of medications when scaling-up interventions.

Results: Funding for the study was secured through a Doctoral Research Award in October 2014 from the International Development Research Centre (IDRC). The study is currently in the data analysis phase and results are expected during the first half of 2016.

Conclusions: This investigation will highlight the detailed processes in place for the care of hypertensive patients in primary health care facilities, and thus also identify the challenges. It will also describe the drug supply chain management systems in place and identify their strengths and weaknesses. The findings, along with the estimates from modeling and simulation, will inform the health system minimum requirements to scale-up interventions to manage and control the hypertension epidemic in the Western Cape province of South Africa.

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KEYWORDS

Hypertension; Manage; Health System; Epidemics; Scaling-up Interventions; Requirements; Supply Chain; Antihypertensive agents; Control; Delivery of Health Care

Introduction

Background and Statement of the Problem

Noncommunicable diseases (NCDs) ‘‘have no borders or boundaries—they are the world’s number one killer and devastate the bottom billion and G20 countries alike’’ [1]. Developing countries are undergoing a process of epidemiological transition from infectious to noncommunicable diseases. The launch of the World Health Organization Action Plan on NCDs, the UN General Assembly Resolution on the Prevention and Control of NCDs, and the Global Alliance for Chronic Diseases (GACD) provide strong evidence that increasing attention is being paid to the impact of noncommunicable diseases on health and development. The United Nations Secretary General Ban Ki-Moon described NCDs as ‘‘a public health emergency in slow motion” [2]. One of the most prevalent NCDs in sub-Saharan Africa (SSA) is hypertension. Hypertension is a complex chronic condition, often referred to as the “silent killer” and a key contributor to the development of cardiovascular and cerebrovascular diseases. Hypertension is often either not diagnosed or untreated, and thus represents a serious unrecognized epidemic in Africa. In 2008, the prevalence of hypertension in SSA was estimated at 16.2%, and it was predicted that in 2025 the figure would rise to 17.4% (95% CI 15.4%-22.6%). As a result, the number of hypertensive patients will increase from 74.7 million in 2008 to 125.5 million in 2025—a 68% increase [3]. A recent systematic review including a meta-analysis predicted the prevalence of hypertension in SSA at mean participant ages of 30, 40, 50, and 60 years to be 16%, 26%, 35%, and 44%, respectively, with a pooled prevalence of 30%. Of those with hypertension, only between 7% and 56% (pooled prevalence 27%; 95% CI 23%-31%) were aware of their hypertensive status before the surveys. Overall, 18% (95% CI 14%-22%) of individuals with hypertension were receiving treatment across the studies, and only 7% (95% CI 5%-8%) had controlled blood pressure. This important and timely study concluded that high prevalence of hypertension, low percentage of awareness, treatment, and control in SSA call for implementation of timely and appropriate strategies for diagnosis, control, and prevention [4].

South Africa, however, has one of the highest rates of hypertension in SSA, where the condition has been well researched compared to other countries in the region. The prevalence of hypertension in the South African population is as high as 44% in adult males living in rural areas. In addition, older adults and males are more affected by hypertension in South Africa [5]. However, unlike other countries in the region, the prevalence has been reported as comparable between rural and urban populations [6], which is attributed to an increase in obesity caused by widespread adoption of Westernized diets throughout the country. The South African National Health and Nutrition Examination Survey (SANHANES-1), released in 2013, revealed that among participants who had undergone a clinical examination and had their blood pressure measured, an increasing percentage of the population had systolic blood pressures that were high (≥ 140 mmHg) in the group 15-25 years of age (5.3%) compared to those 65 years of age and older (63.7%). Half (50.5%) of participants 55-64 years of age had a high systolic blood pressure [7].

Diagnosis, treatment, and control of blood pressure are a major population health problem. Screening campaigns are largely nonexistent because of the generally poor health care infrastructure and low number of health care workers. Most systems cannot afford the additional costs associated with operating a cardiovascular screening program in an already resource-limited environment. Thus, there is an important gap between emerging high-level policies and recommendations, and the near-absence of practical guidance and experience delivering long-term medical care for patients with NCDs within resource-limited health systems.

Purpose of the Study and Significance

To address this gap, our research will consist of field investigations in South Africa to study the implementation and scale-up of diagnosis and drug treatment systems for hypertension. In the context of scaling up access to antihypertensive medications in resource- and infrastructure-limited health care systems, this study will specifically examine the development of an effective drug supply management system and study how to determine the minimum numbers of health care workers, and at what level of training, required at each supply chain interval to ensure adequate care of patients. Our overall goal is to determine the minimum health care system requirements to ensure successful delivery of antihypertensive medications when scaling-up interventions to control the hypertension epidemic in South Africa. The rationale for choosing South Africa for these investigations is that, to our knowledge, it has the most clearly documented hypertension epidemic in SSA, and South African medical care frequently guides other African countries.

Theoretical/Conceptual Framework

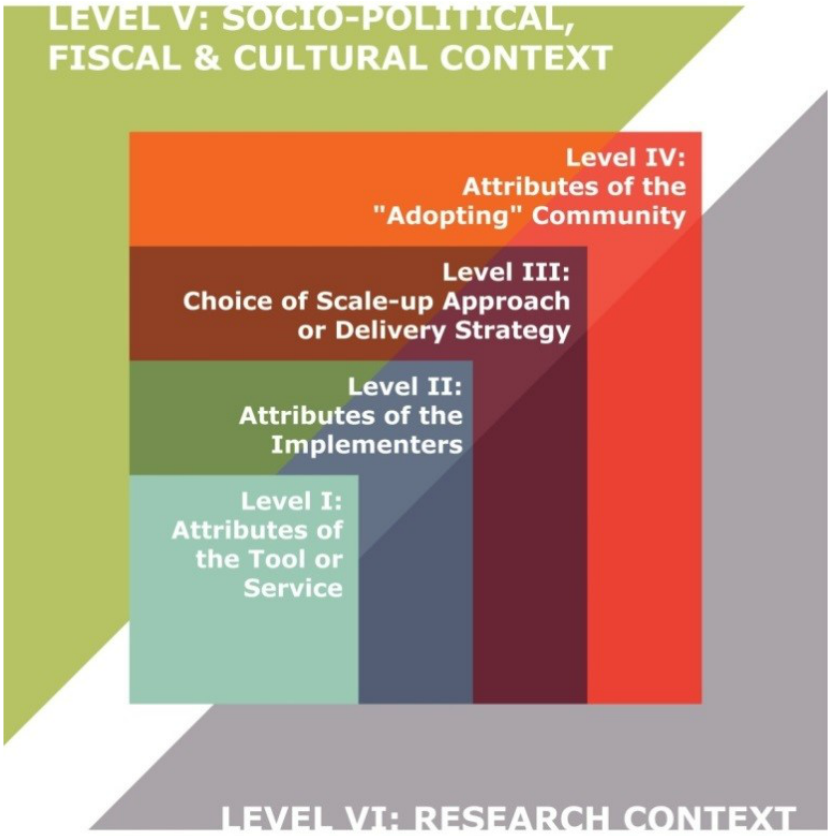
Our focus is on the supply chain as a key element of system responsiveness to hypertension, because to address such an epidemic and reach the maximum number of people, effective and well-structured procedures need to be in place. In addition, a well-functioning supply chain system ensures cost-effectiveness of interventions. In order to be sustained and to expand, a supply chain needs to be robust, agile, and flexible. Systems need to be in place to account for potential reductions in annual funding, increases in drug costs, decreases in drug quality, and loss of drug products. Should any of these situations arise, the management system needs to be designed in such a way that available drugs are equitably distributed to those most in need. Policy makers, program and supply chain managers are under intense pressure to improve health care supply chains to ensure an uninterrupted flow of drugs and related supplies at various service delivery points. They are also under pressure

to produce greater levels of responsiveness, shorter lead times and cycle times for delivery, and positioning of high-quality drug inventory and related commodities to meet the health care needs of millions of people. All these parameters make any scale-up process fairly complex.

Our proposal aims to address these key aspects of the scale-up of an intervention by examining health system requirements to ensure successful delivery of medications to control the hypertension epidemic in South Africa. In order to serve this purpose, we have chosen to use the framework proposed by Gavin Yamey in 2012, hereafter referred to as the "Yamey Framework," illustrated in Figure 1 [8]. We chose this framework because it draws on insights from interviews with scale-up "leaders," many of whom have led national or global health implementation programs, and incorporates themes emerging from relevant recent literature. This framework takes full advantage of the "learning by doing" concept in ways that engage key stakeholders, uses data to address constraints, and incorporates results from pilot projects. Such approaches are very relevant for tackling issues that arise when scaling-up interventions.

The Yamey Framework identifies a range of reported success factors, which are organized into 6 categories, representing different components of the scaling-up process. Inspired by the work of Bergh, van Rooyen, and Pattinson (2008) and Simmons and Shiffman (2007), these categories are the following: attributes of the specific tool or service being scaled-up, attributes of the implementers, chosen delivery strategy, attributes of the "adopting" community, the socio-political context, and the research context [9,10].

Figure 1. Framework for categorizing the study results—The Yamey Framework.



Attributes of the Tool or Service Being Scaled-Up

This framework identifies the importance of simplicity of the intervention and scientifically robust technical policies. Keeping the intervention simple is widely considered to be an important predictor of success. In addition, technical experts who have managed large-scale implementation argue that ensuring technical policies are scientifically robust before going to scale is crucial for success [11].

Attributes of the Implementers

Strong leadership and governance, engaging local implementers and other stakeholders, and using both state and non-state actors as implementers are the key attributes recommended by the framework. These features will be particularly relevant for us in selecting our key informants for the mapping and assessment of the current supply chain management system of antihypertensive medications in South Africa, particularly in the Western Cape region.

Chosen Delivery Strategy

In this category, the framework recommends applying diffusion and social network theories, applying cascade and phased approaches to scale-up, tailoring scale-up to the local situation, decentralizing delivery, and adopting an integrated approach to scale-up. Important lessons from limitations of purely vertical or horizontal interventions prompt more emphasis on integrated approaches.

Attributes of the "Adopting" Community

An engaged, "activated" community is needed. The active participation of the community in planning, implementing, and monitoring interventions is widely cited as a crucial factor in successful scale-up [12]. This is important to avoid blind transfer of solutions from developed to developing countries without integrating local norms.

Socio-Political Context

Political will, national policies, and country leadership are key points. Clear and easy-to-implement policies should be in place to ensure successful scaling-up of the intervention. This aligns well with the concept of a "policy window," which is very important in population health research for the success of interventions, that is, the timing of proposed interventions should be compatible with the political agenda of policy makers. Moreover, health policy research has focused largely on the content of policy, neglecting actors, context, and processes [13]. These four elements should be taken into consideration, and in particular, how these elements interact to shape policy-making. This component of the framework articulates the notion of sustainability, a very important principle of effective population health interventions that we plan to integrate into our research.

Research Context

Incorporating research into implementation ("learning and doing") is the last feature of the framework. We mentioned this earlier as one the main reasons why we chose this framework. Some have argued that successful scale-up "requires the systematic use of evidence to guide the process and incorporate new learning" [10].

Research objectives

Our overall objective in this study is to determine health care system requirements to ensure successful delivery of antihypertensive medications when scaling-up interventions to manage and control the hypertension epidemic in South Africa and the Western Cape Province in particular. No a priori hypotheses have been formulated. Instead, this program of research will use a combination of primary, administrative, and survey data to undertake the necessary analyses to meet the following three specific objectives:

1. To understand the challenges of the health care system in the care of hypertensive patients;

2. To map and assess the current supply chain management system of antihypertensive medications in the Western Cape province of South Africa;

3. Using modeling and simulation tools, to understand how to determine the minimum numbers of health care workers required at each supply chain interval to ensure successful delivery of medications when scaling-up interventions.

Methods

The overarching research question of this thesis proposal is to determine the health care system requirements to ensure successful delivery of antihypertensive medications in the developing country of South Africa when scaling-up interventions to control the epidemic of hypertension. System requirements include materials and human resources linked by processes and policies. This research question will be addressed using a variety of research methods appropriate to each subresearch question presented here, which are informed by the theoretical framework.

Study Design

This combined qualitative and quantitative study will be designed around 3 specific objectives (see Figure 2).

At a very broad level, an antihypertensive medication supply chain will include processes to quantify the drugs needed, procure the drugs, store and control the inventory, distribute the drugs to clinics, and ensure an adequate level of quality assurance. We will conduct field work and have access to secondary data through the South African Medical Research Council (SA MRC). We chose the Western Cape region of South Africa for our field investigations mostly because of the facilitated access through the SA MRC. We recognize that the epidemiology of hypertension may vary from one region to another; however, our research mainly focuses on procedures and operations within the health care system that are comparable between regions. Results obtained in the Western Cape region certainly won't be generalizable throughout the country; however, they could serve as a reference for similar studies in other regions and methods and models could also be replicated.

First, our study will assess the challenges of the health care system, particularly in the Western Cape region, in the care of hypertensive patients. An assessment of standard procedures in a number of health care facilities will identify key processes and individuals' responsibilities in diagnosis and treatment provision. We will combine secondary and primary data to perform this assessment. Primary data will be collected through key informant interviews of medical personnel in the form of semi-structured questionnaires. Data collected will help to describe how patients are processed at the health center, from arrival through health assessments and medication collection to departure, with details on individual processes and health care facility personnel involved. Individual processes for each type of health care facility involved will focus on elaborating current tasks with duration estimation, and identifying potential tasks that could be added or removed to inform task-shifting options. Key informants will be facility managers and health care providers (nurses, pharmacists, doctors, allied healthcare providers) working in health centers of the Western Cape region in South Africa. We will select at least 6 public health care facilities equally distributed between the Cape Town metro (urban) and rural area. The main inclusion criterion for the selection of a health care facility for our study is the implementation of at least a weekly dedicated hypertension or chronic NCD treatment clinic. Secondary data to complement our field investigation, such as administrative hospital data and pharmacy inventory data will be gathered at the health care facilities. Overall, through primary and secondary data analysis, we expect to obtain information on levels of staffing, hypertension screening practice in the health center (routine or opportunistic), volume of patients visits over a certain period such as the last 12 months (% with hypertension), average time

allocated to patient per consultation, presence of task-shifting mechanisms, level of adherence to treatment, availability of drugs at the site and cost to hypertensive patients, frequency of drug shortages, presence of an equitable distribution policy during drug shortages, blood pressure measurement equipment and servicing frequency, and nurse confidence in successfully delivering tasks outside of the traditional scope of attribution in regard to chronic disease management. Data will be analyzed with consideration of sex and gender to determine if there are unique sex and gender differences in the control of hypertension. Data collected in public health centers will be analyzed qualitatively and quantitatively to understand the current state of practice and highlight strengths and challenges of the health care system in regards to hypertension diagnosis and treatment.

Second, the study will investigate the current state of the drug supply chains for antihypertensive medications in South Africa, particularly in the Western Cape region, using an exploratory design. This will be done through mapping of the current public drug supply chains for antihypertensive medications, which will uncover gaps, weaknesses, and strengths of the current supply chain. Practically, we will describe the supply chain by following the flow of medications backward from the service delivery point or health center, to the program management unit where drug selection and quantification take place. This investigation will require interviewing key informants at each step to determine operational procedures, stakeholders, and next steps. Semi-structured interviews will be administered with both closed and a few open-ended questions. Other individuals who will be identified during interviews will also be contacted for their input. In addition, secondary data available at each step will be collected and analyzed to estimate trends, frequencies, uncertainties or fluctuations, lead times, stock shortages, volumes, and potential needs when scaling-up. As we build on existing infrastructure, scaling-up services in this project will involve doing more than what is currently achieved by the health care system in terms of coverage, quality of services, and cost effectiveness of processes. This process will help to identify factors that either enhance or hinder accessibility to antihypertensive drugs for South African population sectors vulnerable to hypertension epidemics. In addition, it will develop a comprehensive supply chain map that will reveal the labyrinth of the South African logistics infrastructure, distribution channels, government regulations, and business customs. The identification of potential leverage points will also inform our strategy in scaling-up interventions to control the epidemic of hypertension. A basic model for logistics process redesign will serve as the basis for describing and analyzing logistics processes involved in the flow of information and goods of the public drug supply chain (see Figure 3) [14].

At the management level, we will select key informants from the provincial department of health (DOH) and use available secondary data to identify procedures in place to gather information estimating how much of each drug is needed. For the procurement level, which pertains to selecting suppliers, placing and monitoring orders, checking delivery quantities and quality, and paying suppliers, key informants at the DOH, the unit in charge of drug supply will be interviewed, combined with available data to identify lead times, ordering frequencies, and demand patterns. Data from this level will also help identify major suppliers and lead times. The distribution component will be investigated by interviewing managers at these levels to gather information on order reception, storage, stock control, transportation, and record keeping for monitoring and control, which will be analyzed to determine shortage frequencies, inventory management practices, stock fluctuations, and uncertainties. The service delivery points will provide data on prescription, dispensing and use of drugs, and patients' compliance with prescriptions. We will complement our investigation by using various data sources from the South African National Health and Nutrition Survey (SANHANES), Hypertension Society of Southern Africa, trade, government reports, manufacturers, and customs to perform our mapping. We expect to identify and evaluate the key elements and processes of the current supply chain management system of antihypertensive medications in South Africa, particularly in the Western Cape region. Special attention will be given to principal public actors (government departments, various levels), volumes of supplies per year, cost of supplies per year, majors suppliers, frequencies of orders, transportation actors and processes, shortage management strategies, variation of demands level, storage conditions and processes, inventory management, quality assurance processes, staffing levels and numbers at each interval, volume level at each storage level, monitoring and record keeping of drug availability, and utilization patterns. We will focus mainly on public sector primary healthcare clinics known as Community Health Centers (CHCs) for this study, as they provide a network of accessible and free primary care for acute and chronic illnesses. We believe that they are fairly representative of clinical practices for the vast majority of hypertensive patients and the SA MRC, which is our host for the study, has agreed to facilitate access to public sector primary health care clinics in the Western Cape region, with which they already have extensive collaboration. This will help uncover the gaps, weaknesses, and strengths of the current public drug supply chain management system and inform the implementation of the model.

Finally, we will explore and use modeling and simulation tools to determine how to estimate the minimum number of health workers required at service delivery points to ensure successful delivery of sufficient medications when scaling-up interventions. In the event that authorities would like to put programs in place to increase diagnosis and treatment of hypertensive patients (ie, scaling-up interventions), modeling and simulation will help to determine what would be needed of the health care system to tackle such a volume of patients. Our study is not actually implementing any scaling-up activity. Data analysis of the earlier study component on how hypertensive patients are processed at health centers, with details on individual processes including task descriptions with time duration estimations, will provide parameters for our modeling and simulations. Tasks and time duration estimations per task will be computed in our modeling to estimate the minimum number of each type of health care worker needed when scaling-up interventions. Quantification of drug needs at service delivery points, which will also be included in the model, will be performed using the World Health Organization's (WHO) "Morbidity Method" [15]. In the standard version of the "Morbidity Method," the total quantity

of drugs required for a health problem is given by the quantity of drugs given as a standard treatment for the problem, multiplied by the number of treatment episodes of that problem. Modeling and simulations will be executed using known hypertension prevalence estimates as inputs. Quantifying the requirement for medication is an essential step in the overall process of medication procurement with the aim of ensuring access. Quantification seeks to answer the question "How much of each medication is needed?" It is the process of determining the quantity of each selected medicinal product required to meet the needs of a specified location (clinic, hospital, region, or country) or program (eg, TB program) for a specified period of time. In addition to being a step in the procurement process, quantification of requirements also provides vital information and a basis for managing the distribution of medication [16].

Figure 2. Linked study objectives.

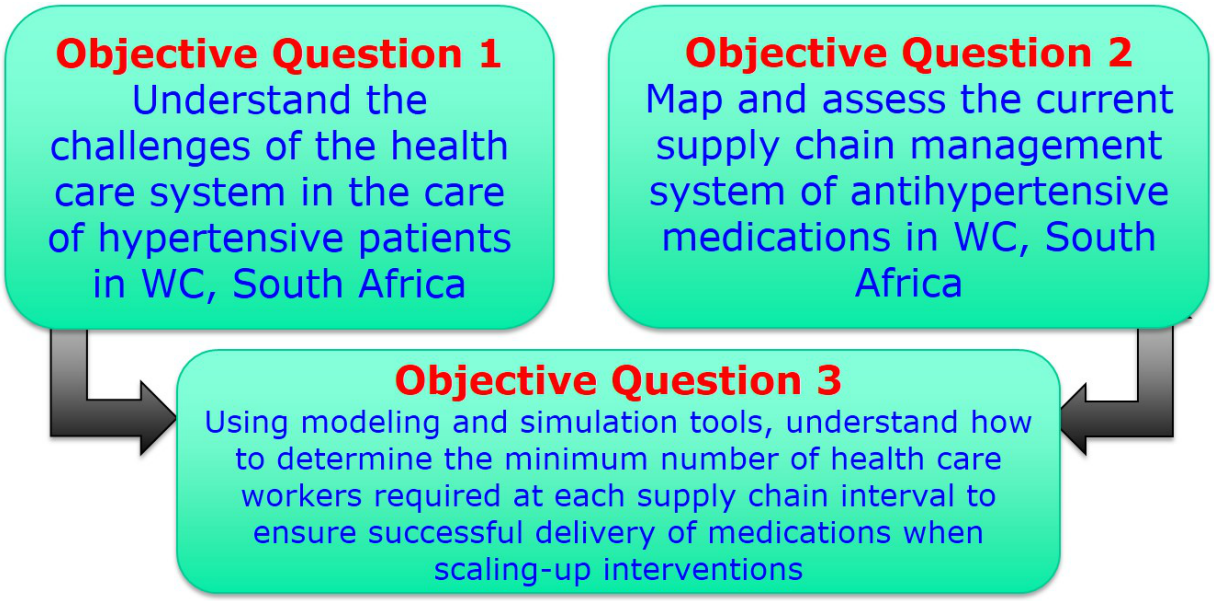
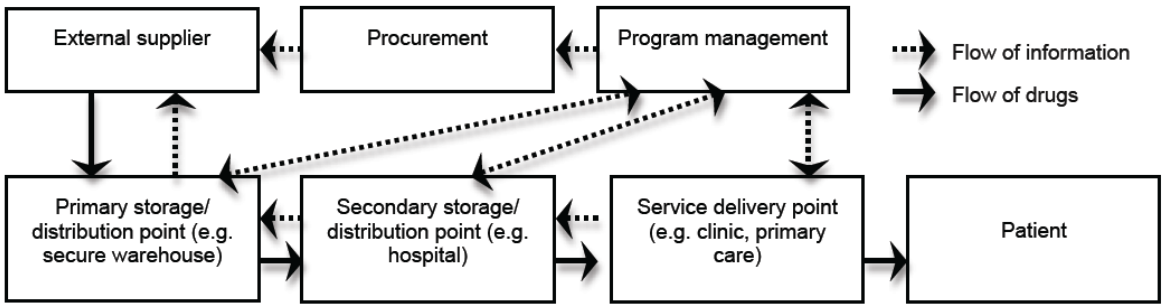


Figure 3. Supply chain management framework (Persson, 1995).



Participants

In this study, our stakeholders are of various types and levels but inform the analysis in a concurrent manner. We will interview only policy makers, administrative staff involved with the drug supply chain, and health care personnel within a few public health centers. No patient will be interviewed as the focus is on processes followed by providers within the health care systems. There will be no translation issues as we expect to use only English when speaking with professional staff. As a general rule, only individuals who have been in their positions for at least 3 months will be included in the study. Each participant will sign a consent form to participate after a verbal briefing about the purpose and structure of the study.

With regard to the mapping of the current public drug supply chain for antihypertensive medications in South Africa, participants will be identified at each stage of the Supply chain management framework. As described earlier in the Study Design section, key informants will be interviewed in project management, procurement, storage, distribution, and service delivery points. This includes but is not limited to key individuals in the Western Cape DOH, pharmacists, and those involved in the procurement, storage, and distribution of medication. Any other organizations or key individuals identified during interviews that may provide useful or additional insight will be asked to participate as well.

With respect to studying how patients are processed at health centers from arrival to departure, with details on individual processes and health care facility personnel involved, participants will be facility managers and health care providers such as nurses, pharmacists, doctors, and allied healthcare providers. This will be an iterative process, such that some key stakeholders may be identified later in the process.

Data Collection Techniques

In this study, the techniques to collect data will be interviews, review of administrative data, and document review. There will be 2 semi-structured questionnaires: one for managers and policymakers in ministries and agencies and the other for medical personnel involved with the hypertension program in selected public hospitals. We will conduct face-to-face interviews; however, in cases for which arranging a meeting will be difficult, telephone interviews will be used as an alternative. We chose to have only open-ended questions in a semi-structured interview format because this gives respondents flexibility to organize and articulate fully their responses [17]. All interviews will be held at the offices of the respondents or at a place and time of their own convenience. At the beginning of every interview, I (the first author) will introduce myself, the purpose of my research, who is funding my research, how I will disseminate the findings of my research, and ask for permission to tape the interview. At the end of the interview, I will ask respondents if they have any comments or questions regarding my research, and also about their perceptions of the current hypertensive drugs supply chain with an emphasis on potential barriers and enablers. This will be helpful in knowing whether I have overlooked critical questions. I will also ask for follow-up interviews to clarify information I do not understand.

Data to inform mapping will be obtained through published survey data and reports, a review of the literature, and interviews with managers and policy makers in ministries and agencies. We will be guided by the elements of the supply chain management framework in the selection of the stakeholders to interview and we also anticipate a "snowball" effect as we progress with our data collection. This is possible because the actual local reality of the supply chain in the country may not completely match our framework. Here, qualitative data will be collected to understand the mechanisms underlying the flow of hypertensive drugs to and within the country, and also to clearly identify all relevant actors. Quantitative data will provide information on volumes, costs and actors as well. A combination of field research, literature review, regulations review, and track record review will be put in place.

Data for the development and testing of the model will be obtained from interviews of medical personnel involved with the hypertension program in selected public hospitals. Secondary data will be obtained from document reviews and administrative databases through the South African Medical Research council regarding hypertension morbidity, demographic health surveys, training records of health care professionals of various levels, numbers of health care workers per level and region, antihypertensive drug consumption, and drug import data. Data from hospital records on screening, follow-up visits, availability of health care personnel, wait times, and daily average numbers of patients received will also be extracted.

All primary data will be obtained through semi-structured questionnaires that will be recorded and transcribed before analysis. Consent will be obtained for recording the interviews and no personal information will be recorded. We plan on obtaining input from knowledgeable individuals of the Western Cape Department of Health to finalize our questionnaires, which can only be done once approval for the study is obtained from the Ethics Committees.

Secondary data will be mostly administrative data and document review. Administrative data will be extracted from accessible surveys such as the South African National Health and Nutrition Examination Survey (SAHANES) and records from health institutions and agencies where we will interview study participants. These data will inform us on the current processes and descriptive statistics will be computed. We will also gather information through document review for the same purposes. Secondary data in this study project will mostly complement primary data gathered through semi-structured interviews. As we approach key informants and health professionals, we will request access to available administrative data and documents. Secondary data will also help us to adapt our semi-structured questionnaire to ask more effective questions according to our research objectives.

Data Analysis

Transcripts will be sent to respondents for their validation, if requested, before data analysis begins. Interview data collected from the field will be coded into 2 basic types: manifest and latent. Manifest coding refers to direct responses to particular questions by respondents. Latent coding considers responses that were not explicitly called for by the question [17]. Themes will be developed out of the interview transcripts and content analysis will also be used to analyze documents. A structured approach guided by the semi-structured interview will be used to determine salient themes emerging from the interviews.

To understand the current state of the drug supply chains for antihypertensive medications in South Africa, data collected through interviews of managers and policy makers in the ministries and agencies will be analyzed to identify social actors, various connections, and processes involved in the drug supply chains for antihypertensive medications in South Africa. These elements will be inputted into the CmapTools software [18] to produce a map organizing and representing knowledge extracted from the data about the current state of the drug supply chains for antihypertensive medications in South Africa. This comprehensive supply chain map will reveal the labyrinth of the South African logistics infrastructure, distribution channels, government regulations, business customs, gaps, weaknesses, and strengths related to the antihypertensive drug supply chain.

Data collected for the development of the model through interviews of medical personnel involved with the hypertension program in selected public hospitals will be analyzed using the qualitative software Atlas ti. We will build a model to determine the necessary drug supply and human resource requirements for the supply chain. Models have proven to be effective forecasting tools for drug supply chains, even in low-resource settings. The model will start at the point of quantification and procurement, and will include the subsequent processes of primary and secondary storage, transportation, and service delivery. The model will also include mechanisms to account for changes in drug demand and will assess different scenarios of procurement, information exchange, and shipment processes [19]. Costs will be integrated at each level of the model to ensure that I (the first author) can create scenarios that are not only

cost-efficient, but effective in ensuring drugs reach the patient in a timely manner. The Supply Chain Intelligence software package available through SAS will be used to execute the model [20]. I expect that this study will determine the minimum number of health care workers, and at what level of training, are required according to population density and size.

We would like to be able to return to our key informants, stakeholders, and participants with our findings and recommendations, not only to disseminate and promote knowledge translation, but also to obtain their feedback and comments that will help to fine-tune, validate, and optimize the mathematical model used.

Results

Funding for the study was secured through a Doctoral Research Award in October 2014 from the International Development Research Centre (IDRC). Approximately 12 months were spent in the field organizing data collection activities, and requesting and obtaining local ethics approval and authorization to access the specific health care facilities and proceed with data collection. Health care providers involved in the care of hypertensive patients were interviewed in 9 primary health care centers (4 rural and 5 urban, around the Western Cape Province) for a total of 54 participants in this part of the study. In addition, 18 key informants at various levels of the pharmaceutical supply chain system in the Western Cape Province were interviewed. Over 55 hours of recordings were collected by myself and then transcribed through a professional third party organization in Cape Town. Supporting documents and other secondary data were also gathered through this exercise. The study is currently at an advanced stage of data analysis and the initial written report is expected during the first half of 2016.

Discussion

Our work will directly strengthen health systems and inform educational programs of the requirements to meet minimum standards. Our program of research will inform policy makers and health care managers in planning and implementing effective health intervention programs that successfully reach all targeted populations, particularly the most vulnerable. This program of research will inform the design of strategies to control and treat hypertension. This will help prevent complications and more importantly reduce the risks of cardiovascular and cerebrovascular diseases, as it is well established that hypertension is a key contributor to their development. As such, our program of research will contribute to the secondary prevention of worsening hypertension and of comorbidities associated with hypertension. Our findings could aid the development of practical guides for delivering long-term medical care for hypertension within weak health care systems characteristic of resource-limited settings such as SSA. Many African countries look towards SSA for infrastructure and guidance when addressing their health system's needs. In addition, policies and recommendations from international organizations could be informed by our research, which will therefore assist with better targeted funding. The possibility of applying our findings to other countries or regions adds great value to our work.

Acknowledgments

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Authors' Contributions

RD developed the initial study protocol. SY and RL reviewed and made substantial contributions to the manuscript, including matters related to conceptualization, discussing, and drafting subsections of the manuscript. All authors reviewed and approved the final manuscript.

Conflicts of Interest

None declared.

Multimedia Appendix 1

IDRC peer-review report.

[PDF File (Adobe PDF File), 457KB - resprot_v5i1e35_app1.pdf]

Multimedia Appendix 2

Confirmation of Defense Thesis Proposal.

[PDF File (Adobe PDF File), 55KB - resprot_v5i1e35_app2.pdf]

Multimedia Appendix 3

Full Ethics Approval University of Ottawa.

[PDF File (Adobe PDF File), 31KB - resprot_v5i1e35_app3.pdf]

Multimedia Appendix 4

Ethics Approval South Africa Medical Research Council.

[PDF File (Adobe PDF File), 179KB - resprot_v5i1e35_app4.pdf]

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Abbreviations

DOH: Department of Health
GACD: Global Alliance for Chronic Disease
MRC: Medical Research Council
NCD: noncommunicable diseases
SAHANES: South African National Health and Nutrition Examination Survey
TB: Tuberculosis
UN: United Nations
WHO: World Health Organization

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Protocol

Predicting Appropriate Admission of Bronchiolitis Patients in the Emergency Department: Rationale and Methods

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Abstract

Background: In young children, bronchiolitis is the most common illness resulting in hospitalization. For children less than age 2, bronchiolitis incurs an annual total inpatient cost of $1.73 billion. Each year in the United States, 287,000 emergency department (ED) visits occur because of bronchiolitis, with a hospital admission rate of 32%-40%. Due to a lack of evidence and objective criteria for managing bronchiolitis, ED disposition decisions (hospital admission or discharge to home) are often made subjectively, resulting in significant practice variation. Studies reviewing admission need suggest that up to 29% of admissions from the ED are unnecessary. About 6% of ED discharges for bronchiolitis result in ED returns with admission. These inappropriate dispositions waste limited health care resources, increase patient and parental distress, expose patients to iatrogenic risks, and worsen outcomes. Existing clinical guidelines for bronchiolitis offer limited improvement in patient outcomes. Methodological shortcomings include that the guidelines provide no specific thresholds for ED decisions to admit or to discharge, have an insufficient level of detail, and do not account for differences in patient and illness characteristics including co-morbidities. Predictive models are frequently used to complement clinical guidelines, reduce practice variation, and improve clinicians' decision making. Used in real time, predictive models can present objective criteria supported by historical data for an individualized disease management plan and guide admission decisions. However, existing predictive models for ED patients with bronchiolitis have limitations, including low accuracy and the assumption that the actual ED disposition decision was appropriate. To date, no operational definition of appropriate admission exists. No model has been built based on appropriate admissions, which include both actual admissions that were necessary and actual ED discharges that were unsafe.

Objective: The goal of this study is to develop a predictive model to guide appropriate hospital admission for ED patients with bronchiolitis.

Methods: This study will: (1) develop an operational definition of appropriate hospital admission for ED patients with bronchiolitis, (2) develop and test the accuracy of a new model to predict appropriate hospital admission for an ED patient with bronchiolitis, and (3) conduct simulations to estimate the impact of using the model on bronchiolitis outcomes.

Results: We are currently extracting administrative and clinical data from the enterprise data warehouse of an integrated health care system. Our goal is to finish this study by the end of 2019.

Conclusions: This study will produce a new predictive model that can be operationalized to guide and improve disposition decisions for ED patients with bronchiolitis. Broad use of the model would reduce iatrogenic risk, patient and parental distress, health care use, and costs and improve outcomes for bronchiolitis patients.

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KEYWORDS

Decision support techniques; forecasting; computer simulation; machine learning

Introduction

Bronchiolitis is inflammation of the bronchioles, the smallest air passages in the lungs, primarily seen in children less than age 2. Within the first year of life, 10% of children are diagnosed with bronchiolitis [1]. By age 2, more than a third of children have had a bronchiolitis diagnosis [2]. Bronchiolitis causes about 71 hospitalizations and 77 emergency department (ED) visits per 1000 infant years [3]. In the United States, each year bronchiolitis incurs around 287,000 ED visits [4], 128,000 hospitalizations [5], and $1.73 billion in total inpatient costs (2009) [5]. For children under age 2, bronchiolitis is the most common cause of hospitalization and represents 16% of all hospitalizations [5-8].

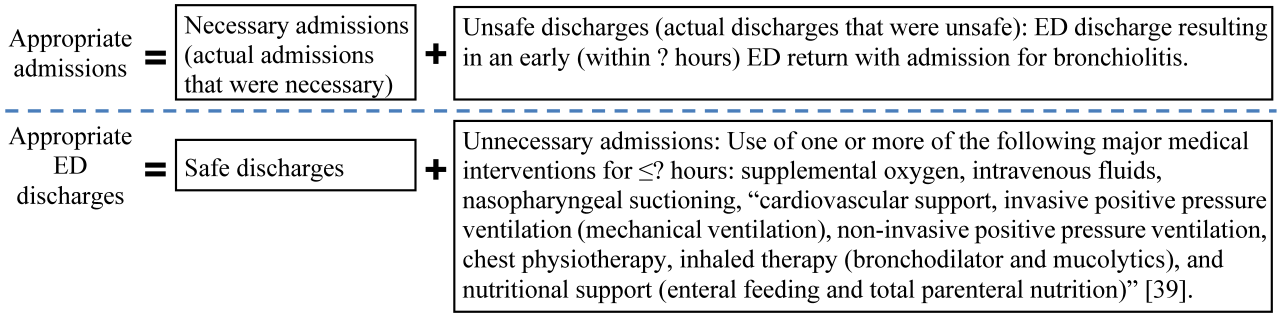
Despite the huge burden of bronchiolitis care, hospitalization decisions are made with insufficient evidence [7,9], resulting in variable admission rates [1,6,9-19]. About 32%-40% of ED patients with bronchiolitis are admitted to the hospital [20-22]. Studies suggest that 20%-29% of these admissions are unnecessary [23,24]. Unnecessary admissions waste health care resources, overwhelm hospital capacity, increase patient and parental distress, introduce iatrogenic risk such as exposure to other infectious diseases, and expose other hospitalized children to the respiratory pathogens of these patients [11,17,25]. As many as 10% of infants affected by bronchiolitis have adverse events while in the hospital [26]. Similarly, about 6% of ED discharges for bronchiolitis are unsafe, resulting in ED return with hospital admission [27] due to inadequate treatment [11]. New approaches are needed to improve ED disposition decision making and reduce unnecessary admissions and unsafe ED discharges.

One method to reduce practice variation and improve clinicians' decision making for bronchiolitis care is to use clinical guidelines [28-31]. However, existing clinical guidelines for bronchiolitis acknowledge that decisions to admit or to discharge are subjective and rely on variable clinical judgment due to a lack of specific objective thresholds with good evidence [30,31]. Clinical guidelines also do not account for differences in patient and illness characteristics including comorbidities [32] and offer limited improvement in determining ED disposition.

Predictive models are an alternative method to reduce practice variation and improve decision making [20-22,33-35]. Predictive models can present objective criteria supported by historical data for an individualized disease management plan. Using data from previous patient encounters to identify patterns, predictive models [36] can overcome the limitations of clinical guidelines. Predictive models can be incorporated into electronic decision-support tools [37] to support the provisional judgment of clinicians or to trigger clinicians to reconsider their judgment in real time [20]. This is especially useful for physicians who see children infrequently or are junior. Typically when results of predictive models are provided, human experts can make better decisions [38].

As reported in our previous paper [39], existing predictive models for ED patients with bronchiolitis are limited by low accuracy and the assumption that actual ED disposition decisions were appropriate. No operational definition of appropriate admission of ED patients with bronchiolitis exists and no model has been built on appropriate admissions (Figure 1). To fill the gap, we will meet the following 3 aims: (1) develop an operational definition of appropriate hospital admission for bronchiolitis, (2) develop a new model to accurately predict appropriate disposition for ED patients with bronchiolitis, and (3) conduct simulations to estimate the impact of using the model on outcomes.

Figure 1. The definition framework of appropriate admission versus appropriate ED discharge that was provided in our previous paper [39]. The details denoted by ""? will be determined by direct evidence in this current study.



Innovations

This study makes the following innovations within the context of bronchiolitis:

1. We will develop a new approach to construct an operational definition of appropriate hospital admission in the ED based on objective data rather than clinical judgment. No such approach currently exists.
2. We will figure out the most important attributes to put into the predictive model using a new simulation method. We will use various attribute combinations to ascertain the minimum requirement on performance and permit tradeoffs for the adaption of our model beyond our setting dependent upon available attributes. Current models are not generalizable beyond the study site because they rely on a

certain set of attributes that may be nonexistent in different electronic medical records.

3. We will build the first model to accurately predict appropriate admission for ED patients with bronchiolitis in real time. No such model currently exists. We will transform bronchiolitis care by developing a predictive model to guide appropriate admission for the first time.
4. Our model will increase prediction accuracy by using a rich set of extracted attributes, including known predictors of hospital admission not used in existing models for ED patients with bronchiolitis.
5. Our model will include environmental variables with a potential for a further increase in accuracy. Air quality environmental variables are associated with the daily number of hospitalizations for bronchiolitis [40] and a child's risk of hospitalization for bronchiolitis within the first year of life [41]. The predictive power of air quality and respiratory virus environmental variables for appropriate admission has never been evaluated.
6. Our study will evaluate the impact of using the model on outcomes. Previous predictive models focused only on accuracy. No impact estimate of using the model on bronchiolitis outcomes has ever been provided.
7. We will use a large data set of 26,701 bronchiolitis patients with high potential to achieve high prediction accuracy. Previous studies are limited by small data sets with typically far fewer than 1000 patients. Many useful predictors of hospital admission cannot be identified in small data sets.

In summary, this study is significant as it will fill gaps by developing a new model to guide and improve disposition decisions for ED patients with bronchiolitis. Broad use of the model will reduce iatrogenic risk, patient and parental distress, health care use and cost, and improve clinical outcomes for bronchiolitis patients. A future study will test the impact of using the model in a randomized controlled trial after implementing the model in an existing electronic medical record to facilitate real-time decision making.

Methods

Machine learning is a field that studies the automatic improvement of computer algorithms with experience. Machine learning methods—such as support vector machine, neural network, and decision tree—are commonly used in predictive modeling [36] and will be adopted in our study. In comparison to statistical methods, machine learning can improve prediction accuracy, occasionally doubling it, with less stringent assumptions on data distribution [38,42,43].

For all 3 aims, we will use the same patient population and data sets:

1. Patient population: Our study cohort includes children under age 2 who had ED encounters at 22 Intermountain Healthcare facilities for bronchiolitis (ICD-9-CM discharge diagnosis code 466.1 [4]) in the past 10 years. Intermountain Healthcare is the biggest health care system in Utah, comprising 185 clinics and 22 hospitals.
2. Data sets: A large administrative and clinical data set in the enterprise data warehouse (EDW) of Intermountain Healthcare will be used. The Intermountain Healthcare EDW contains a vast set of attributes [44]. Our Intermountain Healthcare data analyst will run SQL queries to obtain a data set that has been de-identified and encrypted and then securely transfer it to a computer that is encrypted and password-protected. Secondary analysis will be conducted on the computer. Intermountain Healthcare has dedicated tables to identify changes in procedure and diagnosis codes. The data set contains electronic documentation of about 85% of pediatric care delivered in Utah [45] and includes approximately 400 attributes. A partial list of categories of these attributes includes: admission date and time; age; orders (eg, medications, labs, exams, immunizations, imaging, counseling, etc), including order name, ordering provider, performing date, and result date; allergies; chief complaint; diagnoses; discharge date; exam result; facility seen for the patient visit; gender; health insurance; health care cost (billed charge, Intermountain Healthcare internal cost and reimbursed cost); height; home address; immunizations; lab test result; language(s) spoken; medication refills; primary care physician as listed in the electronic medical record; problem list; procedure date; procedures; provider involved in the visit; race/ethnicity; referrals; religion; visit type (inpatient, outpatient, urgent care, or emergency department); vital signs; and weight [46].

For the last 5 years, data captured cover more than 2900 patients under age 2 and 3500 ED encounters for bronchiolitis per year. Due to its attribute richness and large size, the data set provides many advantages in the exploration of the proposed predictive models. Furthermore, we will use 21 environmental variables that regional monitoring stations recorded over the past decade within the Intermountain Healthcare region. These variables include carbon monoxide, nitrogen dioxide, particulate matter up to 2.5 μm in size and 10 μm in size, ozone, sulfur dioxide, relative humidity, temperature, precipitation, wind speed, dew point, and activities of each of the following viruses: enterovirus; adenovirus; parainfluenza virus types 1, 2, and 3; human metapneumovirus; influenza A and B viruses; rhinovirus; and respiratory syncytial virus. The data for all nonvirus environmental variables came from federal data sources [47,48], which provide such data throughout the United States. Observation unit admissions will be treated as hospital admissions since the only pediatric observation unit within Intermountain Healthcare has the same admission, coding, billing, and documentation requirements. Our analysis will consider various attribute combinations to ascertain the minimum requirement on performance and will permit tradeoffs for the adaption of our model beyond our setting dependent upon available attributes. Our analysis results will serve as the basis for future expansion of our models to other clinical data sets and diseases beyond bronchiolitis.

Aim 1: Develop an Operational Definition of Appropriate Hospital Admission for ED Patients with Bronchiolitis

In a recent paper [39], we provided a definition framework of appropriate hospital admissions. As shown in Figure 1, we

equate appropriate admissions to necessary admissions and unsafe discharges. We equate appropriate ED discharges to safe discharges and unnecessary admissions. The definition uses several threshold values, such as the maximum number of hours for which major medical interventions are used. Using a data-driven approach, we will fill in these values and develop an operational definition to be used in Aims 2 and 3.

For unsafe discharges, we will examine the distribution of the interval between discharge from the ED and a return visit resulting in admission for bronchiolitis within the period of 2 weeks [49,50]. The 95th percentile of the interval will cover most readmissions and define the return threshold for unsafe discharge. The distribution is highly skewed toward a short interval [27]. Thus, the return threshold will be insensitive to the length of the period chosen.

For unnecessary admissions, we will examine the patients who stayed in the hospital for 12 hours or less and were discharged without readmission for bronchiolitis within 2 weeks. These patients are likely to have been admitted unnecessarily. Their median duration of using major medical interventions (Figure 1) will serve as a conservative threshold for use of major medical interventions in all admissions. Unnecessary admissions are those with major medical intervention exposures for no longer than the threshold. We will conduct sensitivity analysis to evaluate the impact of interactions between major medical interventions and other variables.

If the operational definition for all bronchiolitis patients lacks face validity, we will examine data distributions for different age groups to obtain operational definitions by age group. Since the medical interventions for bronchiolitis have not changed over the last 10 years, we would expect the operational definition to remain the same during this period.

Aim 2: Develop and Test the Accuracy of a New Model to Predict Appropriate Hospital Admission for an ED Patient with Bronchiolitis

We will use clinical, administrative, and environmental variable attributes to build machine learning models to predict appropriate hospital admission for individual ED patients with bronchiolitis.

Data Pre-Processing

Traditional techniques like imputation will be used to handle missing values and identify and correct/remove invalid values [36,51]. In the case of environmental variables, classic methods [40,41] will be used to extract aggregate values (eg, daily average) from raw values. In the case of clinical and administrative attributes, grouper models like the diagnostic cost group system will be used to aggregate diseases, drugs, and procedures to reduce attributes [52].

Input Variables

For ED patients with bronchiolitis, predictors of hospital admission have not been exhaustively identified. We compiled in our recent paper [39] a comprehensive list of known predictors. Some of these known predictors (eg, atopic dermatitis [53], low dew point [54], duration of respiratory distress [7], absence of familial atopy [55], enterovirus infection [55], etc) have not been used in existing predictive models for ED patients with bronchiolitis. All known predictors stored in the Intermountain Healthcare EDW and environmental data sets will be used as input variables (ie, independent variables). In addition, our data sets contain attributes beyond known predictors. We will use classic feature selection techniques [56] like the information gain method to find attributes likely to be predictive of appropriate admission. Our team's clinical experts will review attributes, select attributes with face validity, and add these as input variables. With more new predictors of appropriate hospital admission and larger sample size, we anticipate higher prediction accuracy.

Predictive Models

We will use Weka [56] to construct predictive models. Weka is a widely used open-source machine learning toolkit. It integrates a large set of standard machine learning algorithms and feature selection techniques. Both categorical and numerical variables exist in administrative, clinical, and environmental data. Supervised machine learning algorithms that can deal with both categorical and numerical variables, such as *k*-nearest neighbor and random forest, will be used. We will examine each applicable algorithm and tune hyper-parameters manually.

The classic area under the receiver operating characteristic curve (AUROC) [56] performance metric will be used. Our target will be models achieving an AUROC larger than or equal to 0.9, which is considered outstanding discrimination [57]. Some machine learning models, such as decision tree and k-nearest neighbor (ie, similar patients), can be more easily interpreted [58,59]. Other machine learning models, such as random forest, are less straightforward to interpret. If accuracies of models are comparable (AUROC ≥0.9 and ≤0.02 worse for interpretable models compared to less interpretable models), we will favor those that clinicians can more easily interpret.

Sample Size Justification and Performance Evaluation

We have 10 years of data. We will train and test predictive models using a standard method. We will perform stratified 10-fold cross validation [56] on the initial 9 years of data to train predictive models and provide estimates of their accuracy. Data from the tenth year will be used to evaluate performance of the best-performing machine learning algorithm, reflecting use in practice. To figure out the environmental variable, administrative, and clinical attributes necessary for high accuracy, we will use backward elimination [36] to remove input variables so long as the AUROC does not decrease by more than 0.02 or go below 0.9.

No AUROC achieved by current care has been reported before. By extrapolating from statistics reported in the literature (unnecessary admissions up to 29% and unsafe ED discharges of 6%), we anticipate the AUROC achieved by current care to be between 0.6 and 0.8+ [20-24,27]. We will test the hypothesis that the model's prediction will be more accurate by a difference in AUROC of larger than or equal to 0.05. The dependent variable has 2 possible values: appropriate hospital admission and appropriate ED discharge. Assuming a correlation coefficient of 0.6 between the model's prediction result and the actual disposition decision for both values and using a 1-sided

Z-test at a 0.05 significance level, a sample size of 356 instances per possible value of the dependent variable will have 90% power to detect an AUROC increase of 0.05. Data from the tenth year include 3615 ED visits for bronchiolitis, which provides adequate power for testing our hypothesis.

Based on 2 prior studies' results, we anticipate that our model will achieve an AUROC larger than or equal to 0.9 and outperform current care in making disposition decisions. Neither prior study on predicting a bronchiolitis patient's ED disposition is similar to our study, which uses appropriate admission as the gold standard. The first study [21] used actual admission as the gold standard and achieved an AUROC of 0.87. The second study [34] used judgment of an attending pediatrician as well as a length of stay longer than 1 day as the gold standard. The predictive model achieved 81% accuracy, better than an average admitting resident's disposition decision.

For ED patients with bronchiolitis, 17 known predictors of hospital admission (Table 1) are consistently recorded at Intermountain Healthcare facilities and available as structured attributes in our data sets, along with many other potential predictors. We will start building our model using structured attributes. If the model cannot achieve high prediction accuracy, we will extract additional de-identified input variables from ED clinical notes by conducting medical natural language processing on the HIPAA-compliant Homer computer cluster at the University of Utah [60]. For instance, additional input variables include the 8 known predictors of hospital admission (Table 1) that are inconsistently recorded in clinical notes at Intermountain Healthcare facilities.

Table 1. The list of known predictors of hospital admission for ED patients with bronchiolitis recorded at Intermountain Healthcare facilities.

Category	Predictors
The known predictors that are consistently recorded at Intermountain Healthcare facilities and available as structured attributes in our data sets	SpO_2, heart rate, respiratory rate, temperature, age, gender, prior hospitalization, prior intubation, abnormal chest x-ray, low dew point (from the environmental variable data set), rhinovirus infection, coinfection, dehydration, history of bronchopulmonary dysplasia, history of eczema, prematurity, maternal/passive smoking
The known predictor that is rarely recorded as structured attributes at Intermountain Healthcare facilities	enterovirus infection
The known predictors that are inconsistently recorded in clinical notes at Intermountain Healthcare facilities	increased work of breathing, poor feedings, decreased feeding, breastfed, abnormalities on auscultation, retractions, family history of atopy, fewer albuterol in the first hour

If our model still cannot reach high prediction accuracy on the entire group of ED patients with bronchiolitis, we will conduct subanalyses to identify subgroups of ED patients with bronchiolitis on which our model performs well. In this scenario, we will apply our final model only to the identified subgroups of patients. These subgroups are identified by certain characteristics, such as comorbidity, prematurity, age, or ED arrival time (eg, daytime vs night, weekday vs weekend) that are typically independent variables in the original model.

We have large data sets. If scalability is a problem with Weka, a parallel machine learning toolkit like Spark's MLlib [61] will be adopted to develop predictive models on the secure Homer computer cluster [60].

Aim 3: Conduct Simulations to Estimate the Impact of Using the Model on Bronchiolitis Outcomes

We will use a method similar to that in Luo et al [46] to establish the model's utility for future use in clinical practice. More specifically, we will estimate the impact of using the model on bronchiolitis outcomes by applying the model to a retrospective cohort, and determine how the model can be generalized to different sites that collect differing sets of attributes. Our model will be developed using data from Intermountain Healthcare. Our simulations will help determine how to implement the model in other EDs. No prior study has either assessed the impact of using a predictive model on bronchiolitis outcomes or found the set of attributes most essential to generalize the model.

Outcomes

We will assess the outcomes of hospital admission, discharge to home, cost, and ED return. The primary outcome is cost. Other outcomes are indirectly reflected in cost and secondary. Each medical claim is companioned by a billed cost, a reimbursed cost, and an Intermountain Healthcare internal cost [52]. The Intermountain Healthcare internal cost [62] will be used because it is subject to less variation resulting from member cost-sharing [52] and more closely reflects actual cost. To deal with inflation, the medical consumer price index [63] will be used to standardize costs to 2014 US dollars. ED returns will be computed using the time interval defining unsafe discharge.

Estimate a Model's Impact

Given a predictive model and a set of input variables, we will estimate the impact of using the model on each outcome. The same method in Aim 2 will be used to train the model on data from the first 9 years. Data from the tenth year have 4 groups: (1) necessary admissions, (2) unnecessary admissions, (3) unsafe discharges, and (4) safe discharges (Figure 1). For each group, we will obtain prediction results, then estimate the outcome if the model's suggestions were followed. For example, consider necessary admissions. The model will erroneously predict that some of these patients should be discharged. We assume that in clinical application, every such patient will incur an unsafe discharge, an early return visit for bronchiolitis, and a cost equal to unsafe discharges' average cost. The overall estimated outcome is the aggregate of outcome estimates in all 4 groups. Similarly, we can determine the minimum requirement of the model's accuracy for the model to be valuable clinically.

Sensitivity Analysis

Intermountain Healthcare gathers a vast range of attributes. A different hospital may gather a portion of these attributes. To ensure that the model is generalizable, we will examine miscellaneous attribute combinations and estimate the outcomes of bronchiolitis when using the modified model. Our estimate will determine which attributes are important to include. In the case that an important attribute is nonexistent in a given ED, the estimate can advise substitute attributes that have a minor impact on bronchiolitis outcomes.

Our complete model will include as many as 400 attributes. Conducting simulations for each possible combination of the attributes is not realistic because of the exponential growth of the number of combinations. As an alternative, an attribute grouping approach will be used. This approach associates attributes that commonly coexist based upon the judgment of our clinical experts. If an attribute in a group is not recorded by a hospital, related attributes in the group are also likely to be missing, such as attributes from the same lab test panel. Grouping will allow us to create and publish a table that lists the groups of possible attribute combinations, including bronchiolitis outcomes estimated via simulations and the trained parameters of the predictive model. If a hospital shows interest in implementing the model, the table can help assess expected outcomes in their environment, whether additional attributes need to be gathered, and if so, which ones. One row in the table will reflect the attributes in the PHIS+ [64] data model that standardizes administrative and clinical attributes from 6 major US children's hospitals. The model of the row will apply directly to at least these 6 hospitals.

Sample Size Justification and Performance Evaluation

We will test 3 hypotheses: use of our predictive model will be linked to reduced (1) costs, (2) ED returns, and (3) hospital admissions. Due to their skewed distribution, cost data will be log-transformed [52]. The primary hypothesis will be accepted if the model lowers the log cost by at least 10% of its standard deviation. We will use 1-sided paired-sample t-test to assess the log cost difference between the model's prediction result and the actual disposition decision. We will use McNemar's test to assess the difference in ED returns and hospital admissions. A sample size of 857 data instances has 90% power to support the primary hypothesis at a 0.05 significance level. Data from the tenth year include 3615 ED visits for bronchiolitis, which offers sufficient power for testing the primary hypothesis.

If performing simulations on 1 computer is too slow for the numerous combinations of attribute groups, we will conduct parallel simulations on the secure Homer computer cluster [60].

Results

We have secured institutional review board approvals from Intermountain Healthcare and the University of Utah for this study. At present, we are extracting administrative and clinical data from the Intermountain Healthcare EDW. Our goal is to finish this study by the end of 2019.

Discussion

The principle of our approach to developing an operational definition of appropriate hospital admission in the ED is general and can be used for other diseases beyond bronchiolitis. Our simulation method will ascertain how a predictive model can be generalized to different sites collecting various sets of attributes, as well as the group of attributes most essential for generalization. This study will use data from a big health care system with numerous heterogeneous facilities spread across a large area. These facilities include EDs at 22 hospitals, ranging from community metropolitan and rural hospitals attended by general practitioners and family doctors with constrained pediatric resources to tertiary care children's and general hospitals in urban areas attended by subspecialists. Each of these facilities has a differing patient population, scope of services, geographic location, staff composition, and cultural background. This variation creates a realistic situation for identifying factors that are generalizable to other facilities across the United States. One of the models produced during simulation will directly apply to at least 6 large US children's hospitals. Moreover, this study will produce a new modeling strategy that can be generalized to other clinical conditions where decision making is uncertain.

In summary, our work will transform bronchiolitis care by developing a new predictive model to guide appropriate admission for ED patients with bronchiolitis. Broad use of the model will lower health care use and cost and improve clinical outcomes for bronchiolitis patients. We will have a new simulation method to estimate the impact of using a predictive model on outcomes in dissimilar data environments. The method can be useful for implementing other models.

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GL was mainly responsible for the paper. He performed literature review, formed the concept and developed the study, and wrote the paper. BS, MJ, and FN provided feedback on various medical issues, contributed to conceptualizing and designing the study, and revised the paper.

Conflicts of Interest

None declared.

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Abbreviations

AUROC: area under the receiver operating characteristic curve
ED: emergency department
EDW: enterprise data warehouse

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Original Paper

HEART Pathway Accelerated Diagnostic Protocol Implementation: Prospective Pre-Post Interrupted Time Series Design and Methods

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Abstract

Background: Most patients presenting to US Emergency Departments (ED) with chest pain are hospitalized for comprehensive testing. These evaluations cost the US health system >$10 billion annually, but have a diagnostic yield for acute coronary syndrome (ACS) of <10%. The history/ECG/age/risk factors/troponin (HEART) Pathway is an accelerated diagnostic protocol (ADP), designed to improve care for patients with acute chest pain by identifying patients for early ED discharge. Prior efficacy studies demonstrate that the HEART Pathway safely reduces cardiac testing, while maintaining an acceptably low adverse event rate.

Objective: The purpose of this study is to determine the effectiveness of HEART Pathway ADP implementation within a health system.

Methods: This controlled before-after study will accrue adult patients with acute chest pain, but without ST-segment elevation myocardial infarction on electrocardiogram for two years and is expected to include approximately 10,000 patients. Outcomes measures include hospitalization rate, objective cardiac testing rates (stress testing and angiography), length of stay, and rates of recurrent cardiac care for participants.

Results: In pilot data, the HEART Pathway decreased hospitalizations by 21%, decreased hospital length (median of 12 hour reduction), without increasing adverse events or recurrent care. At the writing of this paper, data has been collected on >5000 patient encounters. The HEART Pathway has been fully integrated into health system electronic medical records, providing real-time decision support to our providers.

Conclusions: We hypothesize that the HEART Pathway will safely reduce healthcare utilization. This study could provide a model for delivering high-value care to the 8-10 million US ED patients with acute chest pain each year.

ClinicalTrial: Clinicaltrials.gov NCT02056964; https://clinicaltrials.gov/ct2/show/NCT02056964 (Archived by WebCite at http://www.webcitation.org/6ccajsgyu)

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KEYWORDS

chest pain; decision support technique; interrupted time series analysis; acute coronary syndrome; implementation methods; electronic medical records

Introduction

Background

Current care patterns for acute chest pain fail to focus health system resources, such as hospitalization and cardiac testing, on patients most likely to benefit. Each year, approximately 8-10 million patients complaining of chest pain present to an Emergency Department (ED) in the United States [1]. To avoid missing acute coronary syndrome (ACS), ED providers liberally hospitalize patients with acute chest pain for comprehensive cardiac evaluations (serial cardiac biomarkers and stress testing or angiography). However, <10% of these patients are ultimately diagnosed with ACS [2-6], and this pervasive overtesting costs an estimated US $10-13 billion annually [5,7]. Current guidelines for the management of suspected ACS recommend provocative or anatomic testing as a default strategy, serving to reinforce this overtesting behavior [7].

The Chronic Care Model, as adopted from Wagner's work, identifies the use of evidence-based clinical decision support (CDS) systems as a way to address health system needs and improve health care delivery in chronic conditions such as cardiovascular disease [8-11]. Consistent with this model, accurate ACS risk stratification care pathways are designed to eliminate unnecessary testing and improve quality of care by decreasing false-positive results, nondiagnostic testing, exposure to radiation, and excess costs [12]. The history/ECG/age/risk factors/troponin (HEART) Pathway is an accelerated diagnostic protocol (ADP), which combines a clinical decision aid (the HEART score; Table 1) [13-15], with 2 serial troponin measurements, to identify patients with chest pain who can safely be discharged without objective cardiac testing (stress testing or angiography), either urgently or during follow-up care. Prior studies have established that use of the HEART Pathway reduces cardiac testing by >20%, while maintaining an acceptably low adverse event rate [15-17]. What is needed now is a rigorous evaluation of the implementation of the HEART Pathway into *real-world* clinical settings to determine its effectiveness.

Table 1. The HEART score.

Category	Description	Points
History	Highly suspicious	2
	Moderately suspicious	1
	Slightly suspicious	0
ECG	Significant ST-depression	2
	Nonspecific repolarization abnormality	1
	Normal	0
Age	>65	2
	45-65	1
	<45	0
Risk Factors	3 or more risk factors	2
	1-2 risk factors	1
	No risk factors	0
Troponin	>3x normal limit	2
	1-3x normal limit	1
	<normal limit	0
Total		

Objectives and Hypotheses

In this paper, we describe the rationale and methods utilized to test the effectiveness of the HEART Pathway ADP within a health system consisting of three diverse hospital settings. We hypothesize that implementation of the HEART Pathway will significantly reduce hospitalizations and the rate of objective cardiac testing among low-risk patients, without increasing adverse cardiac events.

Methods

Study Design

This prospective pre-post interrupted time series design compares the risk stratification of patients with acute chest pain before and after implementation of the HEART Pathway ADP. Wake Forest Baptist Health is a three-hospital academic health system located in the Piedmont Region of North Carolina. It

consists of a large quaternary academic medical center with approximately 104,000 ED visits annually, a small community hospital with an annual ED volume of about 37,000 patients, and a free standing ED in an adjacent rural county with approximately 12,000 annual ED visits. This study is approved by the Internal Review Board of the sponsoring organization and is registered with clinicaltrials.gov (NCT02056964).

Eligibility

The target population is adult patients with acute chest pain, in which the provider is concerned about possible ACS, but does not have evidence of an ST-segment elevation myocardial infarction (STEMI) on electrocardiogram (ECG). Therefore, adult patients (aged ≥ 21 years) with acute chest pain, provider-ordered troponins, and without STEMI will be included. Based on STEMI rates at Wake Forest Baptist Health, we expect <5% of patients with acute chest pain to be excluded due to ECG criteria.

Study Timeline

Pre- and post-HEART Pathway ADP implementation cohorts will each accrue patients with acute chest pain for 1 year, with a 3-month wash-in period (Figure 1). Data will be collected electronically from all patients using health system electronic medical records (EMRs), and claims data from insurers will be used on a subset of patients insured by Blue Cross Blue Shield (BCBS) of North Carolina (the largest insurer in the state), MedCost, or North Carolina Medicaid.

Figure 1. HEART Pathway Implementation prospective cohort design.

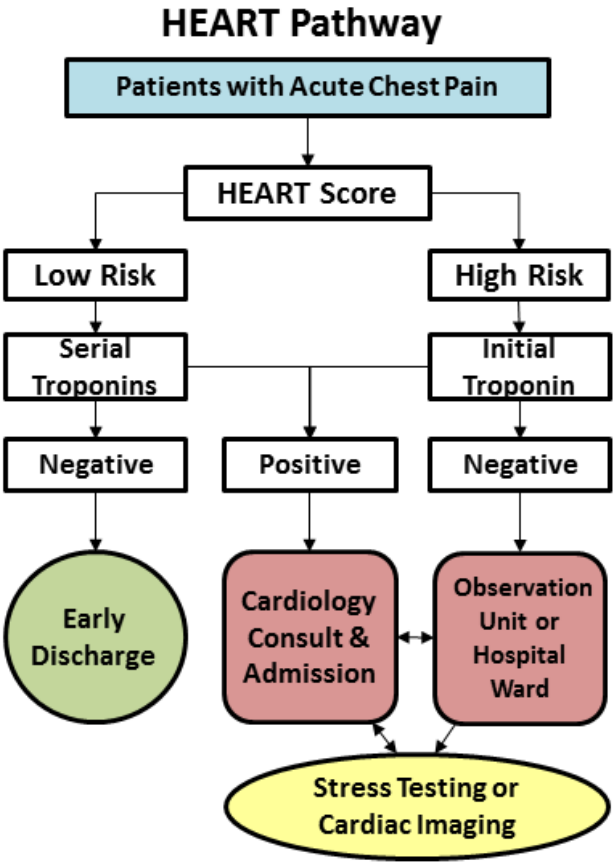
Pre-Cohort: Usual Care	Wash-In	Post-Cohort: HEART Path
1 year	3 months	1 year
~5000 patients	~1250 pts	~5000 patients

HEART Pathway Implementation

The HEART Pathway intervention will incorporate elements of the Chronic Care Model framework (decision support and clinical information systems) by providing test ordering and disposition decision support to ED practitioners and personalized care planning for patients with acute chest pain [8,18,19]. This intervention uses a clinical decision aid (the HEART score; Table 1) [13-15], with 2 serial troponin measurements obtained at 0 and 3 hours after ED presentation. The HEART Pathway algorithm (Figure 2) will be integrated into the EMR system, EPIC (Madison, WI, USA), as an interactive CDS tool. A model window (on-screen pop-up) will display the HEART Pathway tool as a Best Practice Advisory when clinically indicated. The child window will require interaction before the user may return to the parent window in the EMR, but will not force the user to utilize the HEART Pathway tool (the alert will be presented as a *soft-stop*). The modal window will appear when a provider has ordered a troponin test on a patient with a chief complaint of “chest pain” or “heart problem.” The on-screen pop-up will facilitate use of the HEART Pathway CDS tool, by leveraging the normal work-flow of ED providers. For patients presenting with other symptoms that are concerning for ACS (ie, dyspnea, left arm pain, or jaw pain), providers will be encouraged to access the HEART Pathway tool manually. The HEART Pathway tool will be accessible from the EMR’s main menu (the ED Navigator).

Once opened, by pop-up or manually, the HEART Pathway tool will prompt providers to answer a series of questions to determine eligibility and calculate a HEART score on eligible patients. The software will calculate a HEART score based on provider responses and give recommendations on further care based on the HEART Pathway (Figure 2). Patients with a low-risk HEART score (3 or less) and negative troponins at 0 and 3 hours will be identified as a population not requiring objective cardiac testing who can be discharged safely from the ED. Patients deemed at high-risk by the HEART Pathway or with a positive troponin (>99th percentile) will be identified for further testing and/or admission. HEART Pathway use will be tracked using weekly EMR reports. This report identifies all patients who meet inclusion criteria, determines whether the pop-up was activated, and whether the HEART Pathway tool was completed by the ED provider. Noncompliant providers, those that ignore the HEART Pathway pop-up, or those that fail to complete the HEART Pathway decision support tool assessment will receive notification and corrective education via email.

Figure 2. HEART Pathway algorithm.

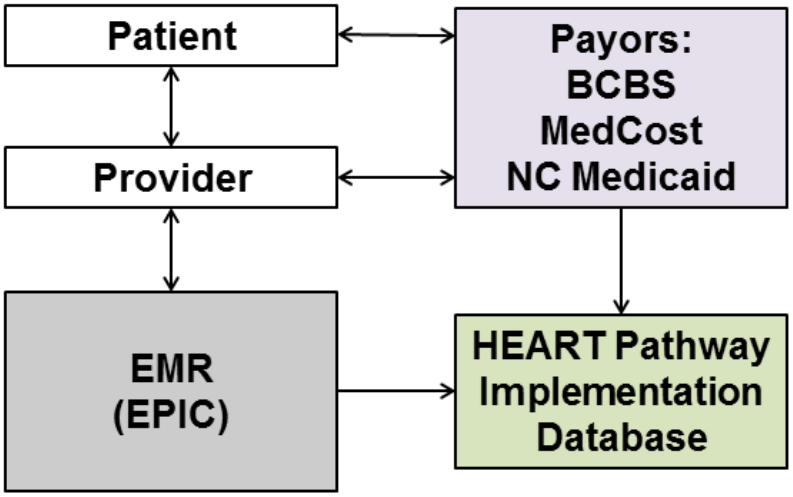


Data Collection

The effectiveness of the HEART Pathway will be assessed using electronic outcome surveillance. EMR data will be collected on all patients with chest pain. Insurance claims data will be collected on patients with Medicaid, MedCost, and BCBS of North Carolina. Data will include patient demographics, past medical history, cardiovascular risk factors, ECG results, troponin results, HEART score and HEART Pathway assessments, ED and discharge diagnoses, disposition, index visit length of stay, objective cardiac testing (stress testing or angiography) at the index visit or within 30 days, recurrent ED visits, readmissions, and myocardial infarction (MI) or death within the follow-up period. Data extraction from the EMR data warehouse (Clarity) will be automated and programmed to pull prespecified data points into our database on patients meeting eligibility criteria on a weekly or monthly basis (Figure 3). Electronic surveillance leverages our informatics strengths and data sharing relationships with insurers, to provide efficient and accurate outcome data. Data accuracy is enhanced by avoiding recall bias inherent in other follow-up methods such as telephone calls. Electronic surveillance also improves feasibility by allowing programmable follow-up on a large number of patients at a low cost. Inclusion of Medicaid patient claims should ensure that our data surveillance includes economically disadvantaged patients and is generalizable to the patient populations of other hospitals. Any discrepancies between sources of data will be adjudicated by study investigators blinded to patient pre- or post-intervention cohort participation. Based on prior studies, we expect 80% of patients to have follow-up data from the EMR or insurer claims. The Social Security Death Master File will be used to search for participants without follow-up data.

The preintervention cohort will be identified using the same inclusion criteria (aged ≥ 21 years with a complaint of chest pain and a troponin ordered without STEMI on ECG) for 1 year prior to integration of the HEART Pathway into cardiovascular care delivery at Wake Forest Baptist Health. The same data elements described above will be abstracted from the EMR and claims databases from BCBS, MedCost, and Medicaid.

Figure 3. Flow of data into the HEART Pathway Implementation Database.



Outcome Measures

Pre- and post-HEART Pathway cohorts will be compared for safety and health care utilization outcomes. The primary outcome will be hospitalization rate at 30 days for patients presenting with acute chest pain. Secondary outcomes will include index hospitalization rate, index and 30-day objective cardiac testing, hospital length of stay (LOS), recurrent ED visits, and nonindex admissions for chest pain. Outcomes will be assessed during the index visit and for 30 days thereafter.

Hospitalization will be defined as an inpatient admission or observation stay. Objective cardiac testing will be defined by any stress testing modality, coronary computed tomography angiography, or invasive coronary angiography. The modalities routinely available at Wake Forest Baptist Health include exercise ECG, exercise stress echocardiogram, dobutamine stress echocardiogram, coronary computed tomography angiography, stress nuclear imaging, stress cardiac magnetic resonance imaging, or invasive coronary angiography. LOS will be the time from ED arrival to hospital discharge for all patients, whether admitted or not. A recurrent visit to the ED will be defined as any patient revisiting the ED with chest pain or other symptoms suggestive of ACS within the 30-day follow-up period.

Safety outcomes will include death and acute MI within 30 days. Missed adverse events will be defined as death or MI occurring in patients discharged from the ED without objective cardiac testing. The definition of MI will be based on the Third Universal Definition of MI, which includes a rising or falling pattern of troponin with a cutoff representing the 99th percentile reference value with a coefficient of variation <10% [20].

Statistical Analyses

Logistic regression models, which include potential patient-level confounders such as age, sex, race, ethnicity, insurance status, and ACS risk factors in addition to the intervention indicator, will be used to assess the effect of the HEART Pathway intervention on hospitalizations (and other dichotomous outcomes such as objective cardiac testing and recurrent ED visits). As secular trends represent a threat to internal validity (observed differences between groups could be from time trends rather than the intervention), we will include the time since the start of the study in the models, as well as the time by intervention interaction. This will allow us to assess the effect of the HEART Pathway implementation on the proportion of patients hospitalized, as well as its effect on the time trend after implementation.

Continuous outcomes will be analyzed using multiple linear regression models including potential confounders, time, the intervention indicator (pre- or post-HEART Pathway implementation), and the time by intervention interaction as described above. LOS tends to be skewed, so we will use some transformation (log, rank, etc) in the analyses if necessary. Residuals will be examined to ensure the model assumptions (linearity, homoscedasticity, and normality) are met. Not all patients will have electronic follow-up data. The default assumption will be that patients without follow-up data from the EMR, insurers, or Social Security Death Masterfile did not suffer adverse events. Sensitivity analyses will be used to assess the impact of missing data on our results. The analyses will be repeated assuming all patients with missing follow-up data had follow-up events (hospitalization, cardiac testing, etc). Multiple imputation will be used to conduct sensitivity analyses that generate complete datasets under a variety of assumptions regarding the rates of outcomes in the two periods. All analyses will be performed using SAS 9.4 (Cary, NC). $P<0.05$ will be considered significant.

Sample Size

A sample size of approximately 10,000 patients, (5000) pre- and (5000) post-intervention, is anticipated. Based on prior studies it is expected that 80% (4000 patients/group) will have follow-up data from the EMR or insurer claims. With this sample size we will be able to estimate safety event rates for each study period to ±0.33% with 95% confidence (assuming an event rate of 1%). Based on our prior studies, we expect a hospitalization rate of 53% and an objective cardiac testing rate of 83% among hospitalized patients during the preintervention period [16,21]. This study will be adequately powered to detect reductions in hospitalization and objective cardiac testing rates

of <5%, with 90% power at the 5% two-sided level of significance. We anticipate larger effects [16,17,22].

Multidisciplinary Collaboration

To facilitate implementation, we have created the Wake Forest HEART Pathway Integration Team consisting of key stakeholders within health system leadership, medical school leadership, and across the disciplines of public health, medical informatics, cardiology, primary care, nursing, and emergency medicine. Through this collaborative effort we will not only engage key stakeholders in the integration of the HEART Pathway with education and care delivery at Wake Forest Baptist Health, but also gain great insights into the potential barriers and facilitators for widespread dissemination and implementation of similar evidenced-based health system quality improvement initiatives across US medical centers.

Results

Preliminary Data

To evaluate the HEART Pathway prior to implementation, we analyzed registry data from 1070 low-risk chest pain patients in an ED-based Observation Unit at Wake Forest Baptist Health. The HEART Pathway identified all 12 patients with major adverse cardiac events at 30 days (100% sensitivity, 95% CI 72-100%) and could have identified 879 of 1070 patients (82%, 95% CI 80-84%) for early discharge without objective testing [16]. Next, we analyzed data from the Myeloperoxidase In the Diagnosis of Acute Coronary Syndromes (MIDAS) Study [23], a multicenter cohort which included 991 participants with suspected ACS from 18 US EDs and data for HEART Pathway risk assessment. ACS was present in 220 of 991 patients (22% of the cohort). In this cohort, the HEART Pathway identified 218 of 220 patients (99% sensitivity, 95% CI 97-100%) with ACS (cardiac death, MI, or unstable angina) within 30 days and identified 200 of 991 patients (20%, 95% CI 18-23%) for early discharge [17]. A lower early discharge rate in MIDAS compared with our first study is explained by the higher prevalence of ACS events in the MIDAS cohort (22% vs 1%). Most importantly, the HEART Pathway had 99% sensitivity in this higher-risk population, suggesting it can be applied broadly to all patients undergoing ACS evaluation. Finally, we conducted the HEART Pathway Randomized Controlled Trial in which 282 adult patients with acute chest pain were randomized to risk stratification via the HEART Pathway ADP or usual care, based on American College of Cardiology/American Heart Association guidelines. In this trial, use of the HEART Pathway decreased objective cardiac testing at 30 days, reduced median LOS by 12 hours (9.9 vs 21.9 hours, P<.01), and increased early discharges. In the usual care group 97 of 141 patients had objective cardiac testing compared with the HEART Pathway group, which had testing in 80 of 141 patients (a difference of 12.1%, 68.8% vs 56.7%, P=.048). In the usual care group, 26 of 141 patients had an early discharge, while 56 of 141 had early discharge in the HEART Pathway group (a difference of 21.3%, 39.7% vs 18.4%, P<.001). No patients identified by the HEART Pathway for early discharge had adverse cardiac events within 30 days and the HEART Pathway was not associated with increased recurrent care [22].

HEART Pathway Implementation

At the writing of this paper, data for this implementation study have been collected on >5000 patient encounters. The HEART Pathway has been fully integrated into health system EMRs, providing real-time decision support to our providers. Data infrastructure has been built such that patients meeting inclusion criteria are included in a registry, including 30-day electronic-surveillance data, which will be integrated with insurer claims data.

Discussion

Study Rationale

This paper describes the design of a prospective cohort study which will determine the effectiveness of the HEART Pathway ADP in safely reducing health care utilization (hospitalizations, objective cardiac testing, hospital length of stay, etc) among ED patients with acute chest pain. This study is timely given the high cost of delivering care to patients with acute chest pain and the current focus on delivering high-value care. The National Quality Strategy, outlined in the Affordable Care Act, focuses on increasing health care quality while simultaneously lowering costs [24]. To adapt to this changing health care landscape, health systems must develop effective methods of translating efficient evidence-based protocols, such as the HEART Pathway, into clinical practice [25-27].

Implementation of the HEART Pathway could improve the value of care for patients with chest pain by decreasing unnecessary health care utilization, false-positive/nondiagnostic testing, radiation, and costs [12]. Our prior analyses of the HEART Pathway, in combination with the HEART Score validation studies [14,15], provide efficacy data on over 7000 patients, and suggest that the HEART Pathway can have a large impact on avoiding testing in low-risk patients, yet retains high sensitivity when applied to higher-risk patients. Furthermore, the HEART Pathway's ability to rapidly identify patients for early discharge and its overall ease of use make adoption in an ED setting feasible. Our experience with the HEART Pathway suggests that patients identified for early discharge will have significant reductions in index LOS and cost [22]. What is needed now is a prospective cohort study, as described in this paper, which will determine the effectiveness of the HEART Pathway by prospectively implementing it in a *real-world* clinical setting.

Nontraditional aspects of this study design include its pre-post interrupted time series design, multidisciplinary team implementation, CDS integrated into the EMR, automated prospective data capture, and electronic surveillance of outcomes (including partnering with insurers for claims data). These features will allow the passive accrual and surveillance of over 10,000 patients. Furthermore, this design will provide a template for others interested in pragmatic testing of care pathways within health systems.

Limitations

The design of this study has some limitations when compared to a traditional randomized design. Secular trends and provider maturation effects are potential threats to the validity of our

pre-post time series design. The electronic surveillance used in this study may increase loss-to-follow-up rates compared with traditional methods of follow-up. However, to determine effectiveness, the HEART Pathway must be utilized in an *all-comers* ED patient population. Randomized clinical trials, due to the consent process, have an inherent selection bias which can threaten the validity of an effectiveness study. Furthermore, given the size and scope of this implementation study, this design is more feasible and cost effective than a clinical trial.

Conclusions

We hypothesize that this study will demonstrate that HEART Pathway implementation within a health system will result in meaningful reductions in health care utilization without compromising patient safety. We expect the HEART Pathway to decrease hospitalizations for comprehensive cardiac evaluations and the rate of objective cardiac testing among patients with a low pretest probability for ACS. In addition, our prior data suggest that the HEART Pathway will shorten hospital LOS for patients with acute chest pain, which should translate into significant cost savings and efficiency gains. Success of this study could provide a model for health systems to provide high-value care to 8-10 million patients who present to US EDs with acute chest pain each year.

Acknowledgments

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Conflicts of Interest

None declared.

Multimedia Appendix 1

Peer Review from AAMC/Donaghue Foundation Capacity-Building Grant Opportunity for Academic Medical Centers.

[PDF File (Adobe PDF File), 182KB - resprot_v5i1e10_app1.pdf]

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Abbreviations

ACS: acute coronary syndrome
ADP: accelerated diagnostic protocol
BCBS: Blue Cross Blue Shield

CDS: clinical decision support
ECG: electrocardiogram
ED: Emergency Department
EMR: electronic medical record
HEART: history/ECG/age/risk factors/troponin
LOS: length of stay
MI: myocardial infarction
MIDAS: Myeloperoxidase In the Diagnosis of Acute Coronary Syndromes
STEMI: ST-segment elevation myocardial infarction

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Protocol

Developing a Tablet-Based Self-Persuasion Intervention Promoting Adolescent HPV Vaccination: Protocol for a Three-Stage Mixed-Methods Study

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Abstract

Background: Human papillomavirus (HPV)-related cancers are a significant burden on the US health care system that can be prevented through adolescent HPV vaccination. Despite guidelines recommending vaccination, coverage among US adolescents is suboptimal particularly among underserved patients (uninsured, low income, racial, and ethnic minorities) seen in safety-net health care settings. Many parents are ambivalent about the vaccine and delay making a decision or talking with a provider about it. Self-persuasion—generating one's own arguments for a health behavior—may be particularly effective for parents who are undecided or not motivated to make a vaccine decision.

Objective: Through a 3-stage mixed-methods protocol, we will identify an optimal and feasible self-persuasion intervention strategy to promote adolescent HPV vaccination in safety-net clinics.

Methods: In Stage 1, we will define content for a tablet-based self-persuasion app by characterizing (1) parents' self-generated arguments through cognitive interviews conducted with parents (n=50) of patients and (2) parent-provider HPV vaccine discussions through audio recordings of clinic visits (n=50). In Stage 2, we will compare the effects of the four self-persuasion intervention conditions that vary by cognitive processing level (parents verbalize vs listen to arguments) and choice of argument topics (parents choose vs are assigned topics) on parental vaccine intentions in a 2 × 2 factorial design randomized controlled trial (n=160). This proof-of-concept trial design will identify which intervention condition is optimal by quantitatively examining basic self-persuasion mechanisms (cognitive processing and choice) and qualitatively exploring parent experiences with intervention tasks. In Stage 3, we will conduct a pilot trial (n=90) in the safety-net clinics to assess feasibility of the optimal intervention condition identified in Stage 2. We will also assess its impact on parent-provider discussions.

Results: This paper describes the study protocol and activities to date. Currently, we have developed the initial prototype of the tablet app for English- and Spanish-speaking populations, and completed Stage 1 data collection.

Conclusions: Our systematic collaboration between basic and applied behavioral scientists accelerates translation of promising basic psychological research into innovative interventions suitable for underserved, safety-net populations. At project's end, we plan to have a feasible and acceptable self-persuasion intervention that can affect key cancer disparities in the United States through prevention of HPV-related cancers.

Trial Registration: ClinicalTrials.gov http://clinicaltrials.gov/ct2/show/NCT02537756 and http://clinicaltrials.gov/ct2/show/NCT02535845 (Archived by WebCite at http://www.webcitation.org/6e5XcOGXz and http://www.webcitation.org/6e5XfHoic, respectively).

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KEYWORDS

adolescents; intervention development; HPV vaccination; self-persuasion

Introduction

Human papillomavirus (HPV) vaccine coverage in the United States is suboptimal (3-dose coverage in 2013 was 38% and 14% for adolescent females and males, respectively), especially among the underserved (uninsured, low-income, and racial and ethnic minorities) [1]. Guidelines recommend administration of the vaccine series to adolescents [2-5], and the Healthy People 2020 goal for 3-dose coverage is 80% [6]. However, many parents are ambivalent and often remain undecided even following a provider recommendation [7-10]. As a result, parents delay or defer making a vaccine decision [8,11]. Undecided parents are a heterogeneous group—some perceive low risk or poor vaccine efficacy, whereas others are concerned about promoting sexual behaviors, unknown side effects, or are simply not motivated [7,9-11]. Most parent-targeted interventions to date focused on reminding parents about adolescent immunizations [12-15] while few have targeted undecided parents and tried to change factors that affect decision making [16,17]. This National Cancer Institute-funded research protocol uses basic and applied social science research to develop a feasible HPV vaccine intervention, based on the principles of self-persuasion, addressing parental motivation and indecision.

Self-persuasion, defined as the process of generating one's own arguments for changing behavior, is an effective approach to influence motivation and behavior. Based in theories of persuasion [18,19] and cognitive dissonance [20,21], basic behavioral research has demonstrated that self-generated arguments are more effective than arguments from an external source [19,21]. Approaches eliciting self-persuasion have improved diverse behaviors including smoking cessation [22], dietary behaviors [23,24], and safer sex practices [20]. Effects of self-persuasion have been shown to persist from 2-3 months [20,23] to 2-3 years [24,25]. Some argue that self-persuasion is the most effective way to change behavior because motivation for change comes from within the individual [21]. Yet, evidence is unclear about the underlying mechanisms that explain *why* self-persuasion is effective. This is particularly true among underserved populations seen in safety-net systems, given that studies to date have been conducted among diverse, but largely well-educated populations [20,22-24].

Generating one's own arguments for changing behavior may be characterized by two processes—choice and deep cognitive processing. First, people choose which arguments, among various alternatives, are most compelling to them. Consistent with the Self-Determination Theory [26], choice elicits motivation for behavior. Across different behaviors, people are more likely to change a behavior when it has been freely chosen [27,28]. Second, people cognitively process self-generated argument content deeply [29]. Consistent with theories of persuasion [18], argument content is more likely to be convincing when processed deeply [19], because it is more accessible in memory [30]. Therefore, we hypothesize that self-persuasion will motivate parents to opt for HPV vaccination *because* they (1) choose arguments that resonate with them, and/or (2) cognitively process the arguments deeply. Examining choice and deep cognitive processing as basic mechanisms of self-persuasion is a novel synthesis of 2 research literatures. Our research protocol uses quantitative and qualitative methods to clarify each mechanism's effects, jointly addressing an important basic science question and how to construct an optimal self-persuasion intervention for underserved populations.

By leveraging people's own arguments for HPV vaccination, self-persuasion may be a more efficient way to deliver personally relevant messages than tailoring or motivational interviewing. Self-generated messages are similar to tailored messages [22], in which experts collect data from each patient to generate customized feedback addressing their unique needs. Tailored messages are effective *because* they are perceived as more personally relevant [31]. However, tailored interventions are time and cost intensive [32]; thus, directing parents to *generate their own* arguments for the vaccine may be a more efficient delivery method. This may be especially true for HPV vaccine decision making because determinants vary across different racial/ethnic groups [33-35]. Similarly, in motivational interviewing, an established clinical approach, providers encourage patients to verbalize arguments for changing their behavior (ie, "patient change talk") [36,37]. Self-persuasion requires fewer trained staff, less time to complete, and may be easier to implement than motivational interviewing. Although some studies applying self-persuasion have asked people to *write* their arguments for the target health behavior [20,22,24], others have had people *verbalize* their arguments [20,24]. We hypothesize that *verbalizing* arguments using a tablet-based app will be an effective and feasible strategy for underserved populations attending safety-net clinics.

A tablet-based, self-persuasion intervention may also be valuable in priming parents to engage in vaccine discussion with their child's provider. This approach may actually prompt parents to generate concerns or arguments *against* the vaccine—a potential

negative effect [22]. However, the process of identifying concerns may also help prepare parents to express and discuss their concerns with the provider [38,39]. By timing the delivery of the tablet-based intervention immediately before seeing a provider, we can examine whether parents are more likely to respond to the provider's cue about the vaccine and whether providers are able to address concerns. Encouraging parent-provider communication is valuable because providers are seen as credible sources of information about immunizations, particularly for underserved populations [38].

In a 3-stage strategy (Figure 1), we are using quantitative and qualitative methods to develop a tablet-based self-persuasion intervention for parents who are undecided about the HPV vaccine and test basic self-persuasion mechanisms through the following aims.

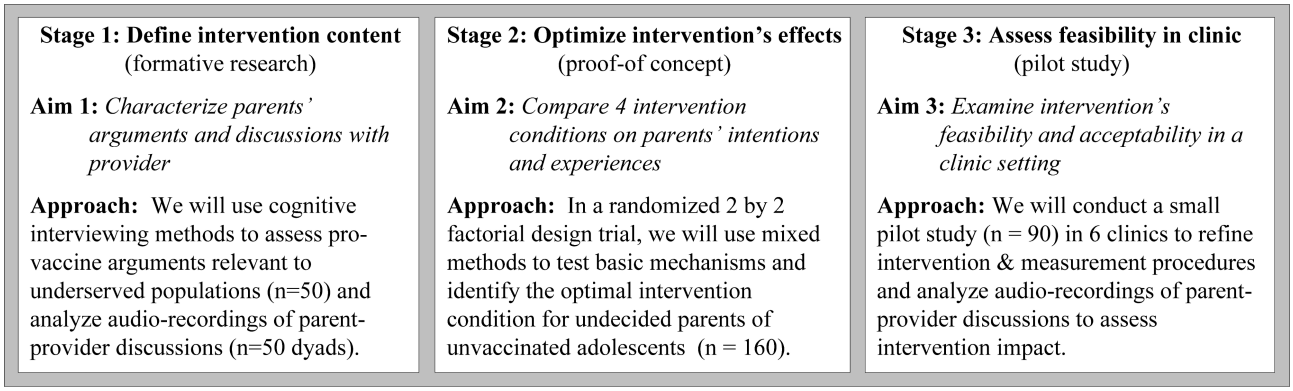
This project innovatively (1) translates basic science findings about self-persuasion into a novel intervention approach to motivate underserved parents to vaccinate their adolescents; (2) elucidates self-persuasion mechanisms that advance basic behavioral science; (3) identifies a more efficient way to elicit similar behavior change effects as tailoring and motivational interviewing; (4) characterizes the communicative environment in which HPV vaccine discussions between parents and providers occur; and (5) uses quantitative and qualitative methods to develop and refine the self-persuasion intervention approach. Our user-centered mixed-methods design synthesizes perspectives from English- and Spanish-speaking families receiving care at safety-net clinics and increases the likelihood that parents will perceive the intervention as relevant [40]. Our systematic collaboration between basic and applied behavioral scientists accelerates translation of promising basic research into innovative interventions suitable for underserved, safety-net populations. At the project's end, we will have a feasible and acceptable self-persuasion intervention that can affect key cancer disparities in the United States through prevention of HPV-related cancers. This paper describes the study protocol and data collection activities to date (currently completed Stage 1).

Methods

Design

We are using a 3-stage mixed-methods design to develop and refine a tablet-based self-persuasion intervention for parents who are undecided about the HPV vaccine (Figure 1).

Figure 1. Three-stage strategy for developing and refining a parent-targeted self-persuasion intervention on adolescent HPV vaccination.



Study Setting

All project activities are being conducted with patients and providers in the Parkland Health & Hospital System. As the integrated safety-net system for Dallas County, one of the largest and ethnically diverse counties in the United States, Parkland's mission is to care for underserved, uninsured Dallas residents. Following the recommendations of the Institute of Medicine, Parkland located 21 school- and neighborhood-based pediatric clinics where there are high numbers of poor and uninsured/underinsured children [41]. Over 14,000 adolescents aged 11-17 years (68% Hispanic, 28% Black, 4% white/other) receive primary care through this system. Parkland's HPV vaccine coverage continues to be lower than national estimates [42,43], but is consistent with clinics serving uninsured, poor populations [44,45]. If this low-rate trend continues, existing HPV-related cancer disparities may widen. Parkland has a standing order immunization policy where providers and nurses recommend all vaccines endorsed by the Advisory Committee on Immunization Practices to unvaccinated patients at all visits. Providers use a comprehensive electronic medical record (EMR) with discrete fields documenting parent refusal and vaccines administered. Parkland participates in the *Vaccines for Children* (VFC) program providing vaccines at free or reduced cost. Thus, this project's parent-targeted self-persuasion intervention complements Parkland's existing infrastructure. We selected the 3 neighborhood- and 3 school-based clinics with the largest volume of adolescent patients aged 11-17 years.

Study Population

For all three stages, eligibility criteria are ascertained by EMR audit. Eligible participants are undecided parents of 11-17-year-old patients who have not started the HPV vaccine series. We specifically focus on undecided parents because they are a large population amenable to self-persuasion effects. Further, our preliminary work with parents decided against HPV vaccine administration suggests that interventions must address their specific worries and concerns. Age is restricted based on guidelines [2-5], eligibility for the VFC program, and parental consent being required for vaccine administration. We are excluding pregnant adolescents due to contraindication and parents who do not provide informed consent, lack telephone access, or have impaired hearing or speech (ie, cannot complete study activities). We ascertain parental indecision for the vaccine

during recruitment. Parents who participate in Stage 1 or 2 will be excluded from later stages. For Stage 1b (audio recordings of parent-provider discussions) and Stage 3 (pilot study), we only select patients with upcoming clinic appointments.

Recruitment

Staff receives weekly EMR reports identifying unvaccinated adolescent patients. Patient information includes name, address, telephone number, birth date, race/ethnicity, language preference, immunization history, and appointment time. Parents are mailed an invitation letter on Parkland letterhead requesting participation in a "project to improve patient satisfaction with health care and delivery of immunizations." The letter provides a telephone number parents can use to opt-out or ask questions. Letters are sent in English and Spanish.

A few days after the mailing, parents who have not refused contact are called by a bilingual research assistant (RA) who explains the project, verifies eligibility, obtains verbal consent, and permission to review their child's EMR, and arranges an in-person study appointment at our research offices or their Parkland clinic. To ascertain eligibility, RAs ask (1) if the child has ever had the HPV vaccine, and (2) what best describes their thoughts about it ("never thought," "undecided," "do not want," or "do want"). Parents who are undecided or never thought about the vaccine are invited and consented. RAs use a computerized database to administer the baseline survey via telephone 5-14 days before the in-person appointment. For parents recruited to Stages 2 or 3, parents are randomized after completion of the baseline survey. Psychosocial variables are assessed again in exit surveys at the end of the study visit to determine changes from baseline. While we do not exclude fathers during recruitment, we expect most participants to be mothers and primary analyses are powered to analyze data from mothers. For Stages 1-3, parents will be given a US $5 gift card for completing the baseline survey and a US $20 gift card after the study visit.

Baseline Survey

On the baseline survey, we assess parent demographics and constructs from health behavior theories or the empiric studies that have demonstrated associations with HPV vaccination behavior. Items and scales were adapted from the published literature and, if not already available, they were translated into Spanish using a multistep process [46,47]. Table 1 describes all baseline measures, estimates of their internal consistency from past studies, and whether they are also assessed during the in-person study appointment for any of the stages. Adolescent demographics, HPV vaccine behavior (date, number of doses, formula), and parental HPV vaccine decision making (acceptance or refusal) are measured via discrete fields in the EMR.

Table 1. Constructs measured at baseline and during study appointments.

Constructs measured at baseline (Cited studies describe survey items and psychometric properties)	Number of Items	Cronbach alpha	Measured during study appointment? (Yes, Stage(s)/No)
Parent demographics: age, race/ethnicity, sex, education, number of children	6	Not applicable	No
General attitudes toward vaccine [48,49]	5		No
Vaccine hesitancy [50,51]	10	.74-.84	No
Knowledge about human papillomavirus (HPV) disease and HPV vaccine [52,53]	7	.70	No
Intentions [54]	3	.96	Yes, Stages 1-3
Precaution Adoption Process Model decision stage [55]	1	Not applicable	Yes, Stages 1-3
Perceived susceptibility [54]	3	.94	Yes, Stages 1-3
Perceived severity [54]	3	.91	Yes, Stages 1-3
Self-efficacy [54]	2	.85	Yes, Stages 1-3
Subjective norms [11,54]	8	.78	No
Perceived benefits [48,56,57]	8	.88	Yes, Stages 1-3
Perceived barriers [54,58-60]	6	.91	Yes, Stages 1-3
Motivation for vaccination [61]	8	.73-.93	Yes, Stages 1a & 2
Trust in provider [62]	1	.86	No
Patient involvement in medical care [63]	18	.83	Yes, Stages 1b & 3

Data Collection Approaches for each Stage

Stage 1

Stage 1 has two components. The goal of Stage 1a is to conduct formative research defining the tablet app's content and creating the four self-persuasion intervention conditions that will be tested in Stage 2. For Stage 1b, the goal is to characterize parent-provider HPV vaccine discussions (Stage 1b).

Stage 1a

We will use cognitive interviewing methodology [64,65] to accomplish the following objectives: (1) develop and refine

question prompts eliciting self-generated arguments, (2) select topics that parents can chose among, and (3) develop peer arguments (Figure 2). We will recruit parents of unvaccinated and vaccinated adolescents (ratio 1:4; *N*=50) to gather the full range of arguments for HPV vaccination. Recruitment will be also stratified on sex of the adolescent.

During the in-person study appointment, the RA will obtain consent and then show the parent how to use the tablet-based app. Voice-over instructions in English or Spanish will ensure literacy level does not inhibit parents' understanding of information presented and tasks of the tablet app.

First, the tablet will guide parents through the deep processing self-persuasion components (Tasks A, C, and D detailed in Figure 2; screenshots of tablet app are shown in Figure 3 with additional examples in Multimedia Appendix 1). In Task A, a short video provides information about HPV infection, related cancers, and the vaccine recommended for both adolescent females and males. In Task C, the tablet audio records parents as they answer a series of assigned question prompts to verbalize pro-vaccine arguments (eg, "In what ways can the HPV vaccine protect your child's health?" or "Some parents mention concerns about the HPV vaccine. What are things doctors or other parents can say to lower parents' concerns?"). Afterward, parents summarize their 3 most important reasons to get the vaccine for their child (Task D). Parents can play back and edit recorded responses until satisfied with them. The RA will use a cognitive interviewing-based guide to probe on comprehension of words, phrases, and vernacular. The RA will mirror participants' own responses one-by-one and ask them to use a Likert scale to rate each question prompt for its difficulty to generate (*not hard at all* to *very hard*) and helpfulness (*not helpful at all* to *very helpful*), as well as open-ended questions to assess preferred prompts (eg, "Which were most clear?"/"Which did you dislike?").

Then, RAs will give the tablet back, ask parents to choose among different HPV topics (Figure 2, Task B), and listen to peer-generated arguments presented in narrative format (Task E). The RA will use an interview guide to assess (1) whether parents can distinguish among the topics, (2) which topics are selected most frequently, (3) whether peer-generated arguments are clear and understandable (eg, "In your own words, what is the main point of this message?"), and (4) if arguments are helpful or raise vaccine concerns.

Figure 2. Stage 1a objectives and Stage 2 trial design with tasks parents complete while using the tablet application.

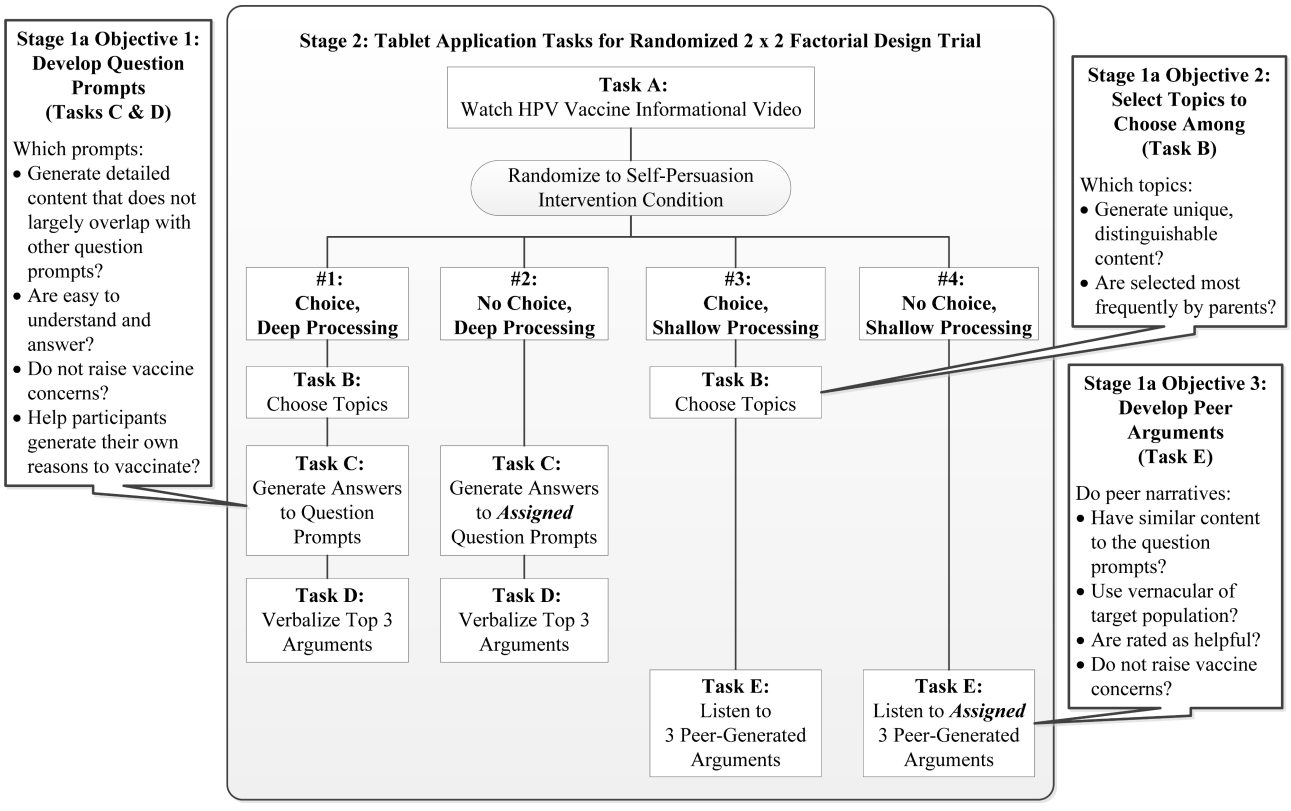
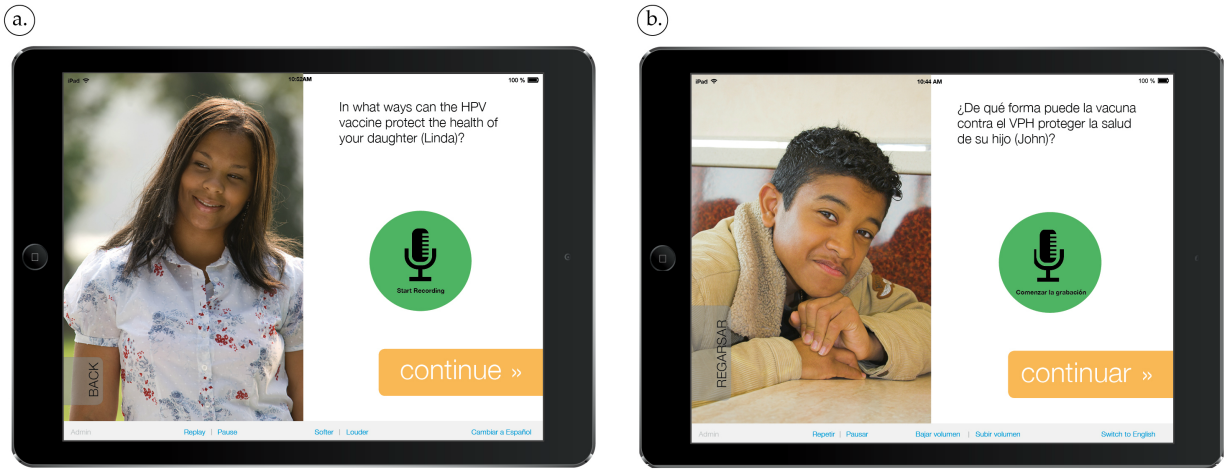


Figure 3. Screenshots of tablet app in (a) English and (b) Spanish.



Analysis Plan for Stage 1a

Cognitive interview data will be transcribed and analyzed through techniques outlined by Willis [64]. We will develop a scheme to code participants' responses to tablet app tasks. Through an iterative process, we will use the codes and participant ratings to examine self- and peer-generated arguments for (1) which question prompts are difficult to answer, (2) which prompts help parents generate their own arguments, (3) distribution of time spent verbalizing each response, (4) distinctions among argument topics, and (5) which argument topics are the most helpful. We will examine data by adolescent sex to ensure we select prompts and topics for Stage 2 relevant for boys and girls.

Stage 1b

The goal is to describe how providers convey HPV vaccine recommendations, how parents express vaccine concerns, and parents' reactions to information from providers. We will identify parents of unvaccinated adolescents with upcoming appointments at the 3 neighborhood and 3 school clinics that see the most adolescents. After using the recruitment strategy described earlier, an RA will meet parents 15 minutes before the clinic appointment to confirm parental consent and obtain the adolescent patient's verbal assent. The RA will place audio-recording equipment in the clinic room. After the participant meets with the provider and is discharged, the RA will conduct a 20-minute exit interview. Parents will be asked open-ended questions about their prior experience with this provider and whether the HPV vaccine was discussed during the visit. Then, the RA will administer a survey to assess household demographics, acculturation, provider recommendation, change in HPV vaccine constructs, and perceived involvement in medical care (Table 1). At least two parent-child dyads per provider will be recruited.

Analysis Plan for Stage 1b

Audio recordings and interviews will be transcribed and analyzed quantitatively and qualitatively.

Quantitative Analysis

We will modify Street's Active Patient Participation Coding scheme, a well-validated observational tool for behavior coding, to analyze the audio recordings [66-68]. For parents, three types of *active* communication will be coded by trained RAs—(1) asking questions, (2) assertive expressions (offering preferences, making a request), and (3) expressing concerns (worries, seeking reassurance). Active "verbalizations" are those that influence discussion content and provider's beliefs and behaviors [68]. Summary scores of the total number in each category per patient per interaction will be generated. Statements to both nurses and physicians are counted, as nurses are often involved in vaccine discussions [69]. For providers, we are assessing (1) vaccine recommendations; (2) rationale for their recommendation including provision of information about benefits and risks; (3) partnership building (open-ended questions encouraging patients to share opinions, feelings, ask questions, and participate in decision making); and (4) supportive talk (verbal statements of reassurance, empathy, or sensitivity) [67].

RAs will co-code 5 recordings to establish rating agreement, code 3 more semi-independently, and discuss coding as a group to resolve differences. To evaluate intercoder reliability, we will use Krippendorff alpha, a measure of agreement that allows for the analysis of categorical and continuous variables in the presence of missing data [70]. We will compute means, standard deviations, and ranges of parent and provider communications. To explore the effect of provider discussions, we will compare parents' responses with baseline and exit survey items (Table 1) regarding the Precaution Adoption Process Model stage of decision making [55], and postvisit perceived involvement in care [63].

Qualitative Analysis

We will perform in-depth thematic analysis of all transcripts using NVivo 9.0. Through iterative coding and interpretation within and across transcripts, a team of bilingual staff trained in qualitative methods will code actual utterances, expressions, and concepts against participant characteristics to identify themes and relationships [71]. We will organize these codes into a codebook that relates data to behavioral theory [72]. Regular meetings will enable the team to test emergent themes and interpretation against the knowledge base of experts in pediatrics, self-persuasion, vaccination, and patient-provider communication.

At the end of Stage 1, we will know (1) the range of provaccine arguments underserved parents generate, (2) which arguments are easiest to generate and most prevalent, (3) which peer-generated arguments are rated as clear, comprehensible, and distinct from other arguments, and (4) range and degree to which parents participate in HPV vaccine discussions. With these data, we will select the optimal argument topics for the self-persuasion intervention conditions. Baseline descriptive information about the communicative environment will be compared with parent-provider discussions after exposure to the optimal self-persuasion intervention in Stage 3.

Stage 2

Stage 2 is a proof-of-concept randomized controlled trial (RCT) in which we will randomly assign 160 undecided parents to one of the four intervention conditions using a 2 (argument topic choice: parents choose vs parents are assigned topics) × 2 (cognitive processing level: parents verbalize vs parents listen to arguments) factorial design (Figure 2). Based on randomization status, the tablet app directs parents to either verbalize their own arguments based on topics they choose (Condition Number 1: deep processing, choice), verbalize arguments based on topics assigned to them (Condition Number 2: deep processing, no choice), listen to arguments based on topics they choose (Condition Number 3: shallow processing, choice), or listen to arguments based on topics assigned to them (Condition Number 4: shallow processing, no choice). Verbally generating (vs reading) material is known to elicit deep cognitive processing [73]. Offering people choice among alternatives has been used to elicit intrinsic motivation for the target behavior [74]. We will use a quantitative approach to test for changes in parents' HPV vaccine intentions and a qualitative approach to compare parents' experiences with intervention tasks to determine which intervention condition is optimal to elicit self-persuasion and minimize negative reactions in our underserved population.

Hypotheses

- Changes in intentions will be higher for deep processing (Conditions 1 and 2) compared with shallow (Conditions 3 and 4) and choice (Conditions 1 and 3) compared with no choice/assigned (Conditions 2 and 4). We are testing two main effects.
- Parents in Conditions 1, 2, and 3 will report experiences with intervention tasks that differ on (1) likeability, (2) usefulness, (3) difficulty, and (4) relevance to discussion with child's provider.

We will identify undecided parents using the same recruitment procedures described above and stratify recruitment based on adolescent sex (80 girls and 80 boys). During a 1-hour study appointment at the child's clinic, parents will complete tablet app tasks depending on randomization status (Figure 2). After using the tablet, an RA will conduct an exit interview to assess parents' perspective on the tablet app, self-persuasion condition, and how their beliefs and experiences shape feelings about the HPV vaccine. The RA will audio record parent responses to the following topics:

- *Quantitative outcomes:* For our *primary outcomes*, we will reassess parents' HPV vaccine intentions and decisional stage. As a *secondary outcome*, we will reassess parents' perceived benefits and barriers. To assess change, we will compare responses to the baseline survey. Using Likert scales, we will ask about parents' experience using the tablet app with respect to (1) likeability, (2) usefulness, (3) difficulty, and (4) relevance for a discussion with their child's provider. Adolescents who accompany their parents to the study appointment can receive the first dose from a Parkland nurse immediately after the exit interview. The nurse will record this dose in the adolescent's EMR to ensure that s/he can complete the series through the VFC program. We will use the EMR to assess administration of all HPV vaccine doses.
- *Qualitative process outcomes:* To determine which conditions are optimal for our underserved population, RAs will observe parents as they use the tablet app and will use open-ended questions to evaluate whether the process raised new vaccine concerns [negative outcome] or addressed concerns [positive outcome].
- *Manipulation checks of choice and cognitive processing*: These checks will provide additional evidence for the hypothesized processes of self-persuasion and help inform which condition we will test in Stage 3.
- *Motivation:* As *a manipulation check of choice*, we will assess motivation for vaccination with a modified Treatment Self-Regulation Questionnaire [61].
- *Memory:* As *a manipulation check of deep cognitive processing*, we will ask parents at the end of the interview to recall as much as they can from the arguments they verbalized/heard [75]. Independent raters will code parents' responses to determine memory accuracy.

Quantitative Analysis

Across self-persuasion conditions, demographic characteristics will be compared at baseline. If intervention groups differ on any of these variables, further analyses will be conducted both with and without these variables as covariates to determine whether these demographic variables are of relevance to group differences. We will compare the effects of choice and cognitive processing on vaccination intentions (primary outcome) using linear regression. Independent variables will be dummy-coded variables based on two main effects (choice: high=0, low=1; processing: deep=0; shallow=1), plus their interaction. If equivalence assumptions of initial scores and parallel regression slopes for the groups are met, baseline intentions will be included as a covariate to properly model change [76]. If not, repeated measures analysis of variance will be used [77]. We anticipate changes in intentions will be highest in the deep processing, choice condition, indicating an additive effect. We will also explore the interaction of the two effects.

Qualitative Data Analysis

We will use the same analytic process described for Stage 1b.

Sample Size

We powered this proof-of-concept trial to test hypothesized effects on a surrogate marker, HPV vaccine intentions. It was

determined by possible effect sizes (f_2) for each main effect, the number of predictor variables for each effect, and the total number of independent variables and covariates in the model (ie, 4, namely, pretest vaccine intention, two main effects, and the interaction). To detect an effect size of f_2=.05, between a small (f_2=.02) and medium effect (f_2=.15), with 80% power and a 5% Type I error rate, we need 160 participants (40 per condition). The sample size will also be sufficient to achieve saturation needed to observe the range of qualitative outcomes.

Synthesis of Quantitative and Qualitative Analyses

To determine which self-persuasion condition (1, 2, or 3) is optimal for our safety-net population and which condition will be tested in Stage 3, we will triangulate quantitative and qualitative findings by creating a summary profile for each condition. The optimal condition will be one that has a positive effect on intentions, but also minimizes participants' negative reactions to using the tablet app. A condition that does not affect intentions will not be considered optimal, regardless of its effect on other quantitative and qualitative outcomes. Likewise, a condition that affects intentions but for many participants raises new concerns, is rated as difficult to complete, or takes significant time to complete will not be considered optimal.

At the end of Stage 2, we will have quantitative and qualitative data that clarify whether it is best to ask parents to verbalize their own arguments, to choose argument topics they prefer, or both. Evidence clarifying which of the two specific mechanisms (deep processing and choice) has an effect, or whether they have an additive effect, will be critical to how we select the optimal self-persuasion condition to implement in Stage 3. For example, if there is an effect of processing but not choice, we would use Condition 2 that has parents verbalize arguments based on assigned topics that are most persuasive rather than allowing them to generate arguments based on chosen topics that may be less persuasive. If there is an effect of choice but not processing, we would use Condition 3 that has parents choose argument topics they want to hear rather than having them go through the more taxing process of verbalizing their own. If both have an effect, Condition 1 will be selected for Stage 3. Given these possibilities of different intervention approaches, our mixed-methods approach maximizes our ability to identify the most optimal intervention condition. Findings will also inform basic behavioral research by generating evidence for specific mechanisms of self-persuasion.

Stage 3

In Stage 3, we will assess feasibility of implementing the optimal intervention, identified in Stage 2, through a pilot RCT with 90 parents in 3 neighborhood and 3 school clinics. Parents will be randomly assigned to either (1) self-persuasion plus information (specific operationalization will be determined in Stage 2; n=45) or (2) HPV information only (n=45). Parents will be asked to come 30 minutes prior to the clinic appointment to meet the RA. For the self-persuasion group, we will follow procedures outlined in Stage 2. The tablet will play the educational video (Figure 2, Task A) to parents in the information-only group. Based on procedures used for Stage 1b, we will audio record the parent-provider discussion. Immediately after the visit, an RA will conduct an exit interview in which participants will be asked questions about whether the tablet app was useful, relevant, culturally appropriate, if they had sufficient time to complete intervention procedures, and their communication with the provider.

Outcomes and Analyses

We designed this pilot RCT to obtain feasibility information on recruitment, clinic implementation issues, and estimation of intervention effects that are key for developing a subsequent efficacy RCT.

Enrollment Rates

We will assess whether enrollment rates are similar across clinics.

Sufficient Time for Intervention Procedures

Because clinics may differ in their patient flow and visit wait times, we will track the number of participants who complete the tablet app within the time constraints allowed by the clinic and determine whether time allotted is similar and sufficient across all sites. We expect an 85-90% completion rate to determine feasibility.

Potential for Contamination

We will determine the appropriate level of randomization (patient, provider, or clinic) and the degree to which contamination occurs at each level. We will use visit history data in the EMR to examine the percentage of patients who visit more than 1 clinic and see more than 1 provider. For example, if there is significant crossover of patients to different providers at the same clinic, then we will randomize at the clinic-level in the subsequent efficacy trial.

Intermediate Outcome: Active Parent Participation

We hypothesize that exposure to the self-persuasion intervention will positively influence active parent participation in discussions with providers. We will apply Street's scheme to code the 3 types of active communication (Stage 1b) [67]. To estimate effect sizes for the subsequent efficacy RCT, we will compute means, standard deviations, and ranges of parent and provider communications and compare them to data collected in Stage 1b. We will use multivariable mixed linear regression modeling to explore factors associated with parent degree of participation (eg, English vs Spanish language) [78]. This method models the provider as a random effect to adjust for potential clustering of patients by provider; parent/patient characteristics of interest will be modeled as fixed effects.

Primary, Quantitative Outcome: HPV Vaccine Uptake

We hypothesize that exposure to the self-persuasion intervention will increase 1-dose and 3-dose HPV vaccine coverage rates. We will use the EMR to measure vaccine uptake. These data will help estimate intervention effect sizes of the self-persuasion intervention, compared with the information-only group, guiding the design and sample size for the subsequent efficacy RCT. We will also measure HPV vaccine-specific measures of intentions, benefits, and barriers (Table 1).

Sample Size

To estimate the sample size necessary to establish feasibility, we used a confidence interval approach and formula for

obtaining a 95% CI for a single proportion. Assuming a priori criterion of success if 1-dose coverage is 70% or more of eligible adolescents and a margin of error of 0.05, the required sample size would be at least 90 patients.

After completing Stage 3, we will have quantitative and qualitative data to determine whether our self-persuasion intervention is feasible and acceptable across clinics—data that will guide us in refining intervention and measurement procedures. Thus, at the end of this stage, we will have a well-characterized and feasible intervention promoting HPV vaccination ready to be tested in future efficacy trial.

Results

Initial Prototype of the App

To date, we have developed the initial prototype of the tablet app and completed Stage 1. Here we summarize the following aspects of the tablet app design to ensure cultural appropriateness for our diverse, low-literacy study population: (1) content of the educational video, (2) conceptual equivalence of content for English- and Spanish-speaking parents, and (3) relevance and appeal to parents of adolescent boys and girls.

Educational Video Content

Educational content was derived from published sources and previously tested educational materials adapted to a 6th grade reading level for low literacy populations [79,80]. We designed messages to address constructs (perceived risk, perceived benefits, perceived barriers [safety and side effects], and anticipated regret) important to our safety-net population based on our formative research and the empiric literature [43,81-83]. The goal of the video was to provide basic vaccine-related facts so that all parents would have the same baseline knowledge of the HPV vaccine prior to completing self-persuasion intervention tasks (Figure 2, Task A). The educational video content was written by co-investigators, translated into Spanish through a multistep process by a bilingual committee (detailed in the following section), and reviewed by our community advisory board (CAB).

We convened 3 CAB meetings during development of the educational video, 2 in English and 1 in Spanish. The sixteen CAB members included social workers who specialize in medical, immigration, and children's services; parents of adolescents; clinic administrators and medical staff; outreach workers; health educators and translators; and community program directors. Each meeting was conducted with at least two research staff to facilitate the discussion and take notes. CAB members each used the iPad independently at the beginning of the meeting to enable detailed feedback and discussion. Members suggested changes to the content and format of the educational video, including facts about the sexual activities that lead to HPV transmission, neutral (nongraphic) images of HPV and its effects on the human body, the pace of the voice-over narration, and text font size. Members at the Spanish CAB meeting stressed the importance of maintaining community trust by giving parents unbiased facts and suggested language to increase parents awareness of what the vaccine does and does not protect against.

Conceptual Equivalence of Study Materials for English- and Spanish-Speaking Parents

All materials (invitation letter, surveys, and tablet app content) went through a multistep translation process in which materials were translated into Spanish, back-translated, tested using cognitive interviewing methods with the target population, and reviewed by a bilingual committee representing several Latin American countries [46,65]. The goal of the translation process was to create conceptually equivalent materials for both English and Spanish speakers and to strive for "broadcast" Spanish (eg, understandable to immigrants from all Spanish-speaking countries) [47,84,85]. The 6-member committee identified potentially problematic concepts (eg, higher literacy phrases and grammar). They designed a cognitive interviewing guide to probe for problems with comprehension and cultural appropriateness.

To accomplish this goal, the committee had to decide when words and phrases should differ between the English and Spanish versions. For example, the phrase "It eases my mind to know the vaccine was carefully tested" was translated as "Me tranquiliza (feel calm) saber que la vacuna fue cuidadosamente probada" to facilitate comprehension of the emotion. In addition, when translating the phrase "better prevent now, than regret later" into Spanish, the words "now" and "later" were dropped to use a well-known phrase in Spanish—"Más vale prevenir que lamenter (ie, better to prevent than to lament)." After cognitive testing in both languages, the committee sometimes identified that the best solution was to change the English text. For example, "chance/oportunidad" was systematically changed to "risk/riesgo." The concept "chance" is appealing from a literacy perspective; however, committee members argued that it did not fully convey the potential for an adverse consequence. Screenshots from the English and Spanish versions of the tablet app are shown in Figure 3 and Multimedia Appendix 1.

Relevance for Parents of Adolescent Boys and Girls

Based on formative research findings in the HPV vaccine literature [82,83,86], the tablet app was targeted to the sex of the child in two ways: gender of the narrator and predominant images selected. Investigators and staff met thrice to evaluate potential male and female narrators for each language and made a final selection based on consensus. Narrators were evaluated based on accent, pitch, and pace that would appeal to parents in our geographic region. The images of the children, single-parent, and two-parent families reflected the racial and ethnic distribution of our target population (see Figure 3 and Multimedia Appendix 1). While both the English and Spanish versions depicted African American, Latino, and white families, a larger proportion of African American images were selected for the English version of the app and Latino images for the Spanish version.

CAB members' feedback on narrators and images were positive overall. Members remarked that the images were visually appealing, but requested more images of boys and a broader range of skin tones for the African American images. CAB members felt that it was important to maintain gender concordance of child and narrator for parents, and appreciated that the audio and text always matched. Bilingual CAB members

were asked to test both the English and Spanish versions of the app and compare their experiences; they reported that the voice-over narration was clear in tone, had a good pace, and would be understandable to parents from any Spanish-speaking country.

Discussion

Vaccine Coverage in the United States

HPV vaccine coverage among US adolescents is suboptimal and interventions that address parental decision making are urgently needed. Self-persuasion—generating one's own arguments for a health behavior—may be particularly effective for parents who are undecided or not motivated to make a vaccine decision. Through a three-stage design, we will identify an optimal and feasible self-persuasion intervention strategy to promote adolescent HPV vaccination in safety-net clinics.

There are some study design limitations that warrant mention. First, in our 2 × 2 factorial trial (Stage 2), we opted not to include 1-dose coverage as an outcome. Given that the purpose of Stage 2 is to understand basic mechanisms and refine and optimize the intervention by examining individual components of it, we opted to conduct the study in a more controlled setting than a clinic visit. As a result, unless adolescents accompany parents to study appointment (which is not required for participation) as they would to a clinic visit, we will be unable to assess vaccine uptake. Instead, we opted to assess vaccine intentions as the primary outcome because meta-analytic evidence suggests that experimentally induced changes in behavioral intentions lead to subsequent changes in behavior [87]. Moreover, we will assess 1-dose and 3-dose coverage (Stage 3), so we will have evidence for the intervention effect on vaccine behavior that will be critical for designing a future efficacy trial. Second, the studies across the three stages are not sufficiently powered to definitively examine potential race/ethnicity and sex differences in the intervention. This is important given that factors influencing parental motivation may differ depending on ethnic/cultural background and whether the child is a girl or boy [43]. However, we will be able to *explore* these potential differences in this study to generate preliminary data about variables that moderate the self-persuasion intervention's effect and thus consider powering the future efficacy trial to test potential moderators. Third, we did not include question prompts to directly rebut vaccine concerns and we excluded parents who were decided against the HPV vaccine. In our preliminary work, we have found that prompting parents to think about vaccine concerns can raise concerns that they were not thinking about without prompting, and persuade parents against vaccination. We believe a separate intervention approach focused on addressing worry and concerns is warranted for these "decided against" parents; thus, it is best addressed in a separate study.

Conclusions

This project's findings will inform basic research by testing specific theoretical mechanisms underlying self-persuasion and providing evidence to support and guide future basic research in self-persuasion. It addresses underserved populations (uninsured, poor, racial, and ethnic minorities) who have high incidence and mortality from HPV-related cancers. The project will enhance the capability of safety-net clinics to promote HPV vaccination by developing a self-persuasion intervention addressing parental indecision. Our three-stage intervention development strategy takes several steps to ensure the usability and cultural appropriateness of all project materials for underserved populations. We are leveraging Parkland's existing EMR to identify eligible patients and evaluate the intervention's impact on HPV vaccine uptake. Our intervention approach holds promise to be institutionalized by Parkland, adapted for other cancer prevention behaviors (eg, smoking cessation, physical activity), and adopted by similar safety-net systems if shown effective in the future efficacy trial.

Acknowledgments

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Conflicts of Interest

None declared.

Multimedia Appendix 1

English and Spanish screenshots depict each of the tasks parents complete while using the tablet application.

[PDF File (Adobe PDF File), 959KB - resprot_v5i1e19_app1.pdf]

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Abbreviations

CAB: community advisory board
EMR: electronic medical record
HPV: human papillomavirus
RA: research assistant
RCT: randomized controlled trial
VFC: Vaccines for Children program

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Original Paper

See Me Smoke-Free: Protocol for a Research Study to Develop and Test the Feasibility of an mHealth App for Women to Address Smoking, Diet, and Physical Activity

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Abstract

Background: This paper presents the protocol for an ongoing research study to develop and test the feasibility of a multi-behavioral mHealth app. Approximately 27 million women smoke in the US, and more than 180,000 women die of illnesses linked to smoking annually. Women report greater difficulties quitting smoking. Concerns about weight gain, negative body image, and low self-efficacy may be key factors affecting smoking cessation among women. Recent studies suggest that a multi-behavioral approach, including diet and physical activity, may be more effective at helping women quit. Guided imagery has been successfully used to address body image concerns and self-efficacy in our 3 target behaviors—exercise, diet and smoking cessation. However, it has not been used simultaneously for smoking, diet, and exercise behavior in a single intervention. While imagery is an effective therapeutic tool for behavior change, the mode of delivery has generally been in person, which limits reach. mHealth apps delivered via smart phones offer a unique channel through which to distribute imagery-based interventions.

Objective: The objective of our study is to evaluate the feasibility of an mHealth app for women designed to simultaneously address smoking, diet, and physical activity behaviors. The objectives are supported by three specific aims: (1) develop guided imagery content, user interface, and resources to reduce weight concern, and increase body image and self-efficacy for behavior change among women smokers, (2) program a prototype of the app that contains all the necessary elements of text, graphics, multimedia and interactive features, and (3) evaluate the feasibility, acceptability, and preliminary efficacy of the app with women smokers.

Methods: We created the program content and designed the prototype application for use on the Android platform in collaboration with 9 participants in multiple focus groups and in-depth interviews. We programmed and tested the application's usability with 6 participants in preparation for an open, pre- and posttest trial. Currently, we are testing the feasibility and acceptability of the application, evaluating the relationship of program use to tobacco cessation, dietary behaviors, and physical activity, and assessing consumer satisfaction with approximately 70 women smokers with Android-based smart phones.

Results: The study was started January 1, 2014. The app was launched and feasibility testing began in April 1, 2015. Participants were enrolled from April 1-June 30, 2015. During that time, the app was downloaded over 350 times using no paid advertising. Participants were required to use the app "most days" for 30 days or they would be dropped from the study. We enrolled 151 participants. Of those, 78 were dropped or withdrew from the study, leaving 73 participants. We have completed the 30-day

assessment, with a 92% response rate. The 90-day assessment is ongoing. During the final phase of the study, we will be conducting data analyses and disseminating study findings via presentations and publications. Feasibility will be demonstrated by successful participant retention and a high level of app use. We will examine individual metrics (eg, duration of use, number of screens viewed, change in usage patterns over time) and engagement with interactive activities (eg, activity tracking).

Conclusions: We will aggregate these data into composite exposure scores that combine number of visits and overall duration to calculate correlations between outcome and measures of program exposure and engagement. Finally, we will compare app use between participants and non-participants using Google Analytics.

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KEYWORDS

smoking cessation, mhealth, diet, exercise, imagery, cell phone, handheld app

Introduction

This paper presents the protocol for an ongoing research study to develop and test the feasibility of a multi-behavioral mobile health app (mHealth app), See Me Smoke-Free, aimed at helping women smokers to quit. Each year, about 180,000 women in the U.S. die from smoking-related illnesses [1]. In 1987, lung cancer surpassed breast cancer as the leading cause of cancer death among U.S. women [2]. Lung cancer causes as many deaths as breast and gynecological cancers combined [2]. Smoking causes about 80% of Chronic Obstructive Pulmonary Disease (COPD) deaths among women annually, and women smokers are more than 10 times more likely to die from COPD than nonsmokers [3]. Women who smoke double their risk of coronary heart disease, and are at increased risk for developing cancer and fractures [1]. Almost 46% of women smokers try to quit annually [4]. The Lung Health Study found that women may benefit from quitting more than men [5].

Special Issues Related to Quitting Among Women

A large body of empirical evidence suggests that women are less successful at quitting smoking [6] and are more likely to relapse than men [7]. This may be because of concerns about gaining weight, lack of social support, stress and depressed mood [8], and physiological symptoms related to the menstrual cycle [6,7,9-11]. Biology may also be a factor as women report greater withdrawal symptoms when they quit smoking than men [12-14], and withdrawal symptoms may be affected by menstruation [11,15,16]. In addition, research indicates that nicotine replacement therapy may be less effective for women [9,17-19].

Women gain 8-10 pounds on average after quitting smoking [20-22]. About 25% of female quitters gain <5 pounds, 50% gain 5-15 pounds, and 25% gain >15 pounds [23-25]. Thus, it is no surprise that half of women smokers report concerns about weight gain as a barrier to quitting [6,26], and most women quitters who relapse identify weight gain as a factor [7]. About half of women smokers report weight control as a reason for smoking [27], and 52% identify weight gain as the reason for relapse [28]. Weight concerns cross age and racial boundaries, but younger, Caucasian women report the most weight concern [29,30]. Two meta-analyses indicated that pre-smoking cessation concerns about weight gain and pre-cessation BMI (Body Mass Index) did not appear to reduce the amount of weight gained when quitting [31,32].

Women who are highly concerned about their weight are less likely to contemplate quitting, and are more likely to smoke as a means to control weight [26]. Several studies suggest that women smokers endorse a thinner body image and are less satisfied with their bodies than never-smokers [33,34]. Weight-concerned women report overall body image as a factor in deciding whether to quit, and in relapse after quitting [26]. A recent meta-analysis suggests that incorporating a personalized weight management program may reduce weight gain without undermining cessation, and recommended further research in this area [32]. Incorporating a body image component into a cessation intervention for women is warranted. Studies have shown that these smokers may be more successful in quitting if they can attain a more realistic body image [34-36].

Addressing Multiple Risk Factors

A small but growing body of literature suggests that a simultaneous, rather than sequential, approach may be more successful for decreasing weight and sedentary activity, and may contribute to successful smoking cessation among women [32,37-39]. According to a recent meta-analysis by Spring and colleagues [38] that included 10 randomized controlled trials, combined smoking plus weight treatment produces significantly higher short-term abstinence and significantly lower weight gain than did smoking treatment alone. The studies included in this meta-analysis showed continued advantage to the multi-behavioral approach on abstinence after 6 months, although the effect on weight control was no longer significant [38]. Other smoking cessation interventions have shown that participation in structured exercise can be an effective smoking cessation strategy compared to general wellness (control) groups although mixed results have been observed [40-42]. While these studies did not target body image per se, meta-analytic results have shown that exercise does improve body image irrespective of changes in fitness or body weight [43]. Thus, the inclusion of exercise behavior as part of a cognitive intervention that targets body image concerns may be an effective way to increase smoking abstinence in women who smoke [44-48].

Theoretical Framework and the Use of Guided Imagery

Body Image

Cognitive behavioral theory (CBT) predicts that cognitive biases lead to overvaluation of body size and shape, internalization of thin body ideals, body image dissatisfaction, and maladaptive weight control practices among women (eg, smoking, dieting)

[49]. Compared to nonsmokers, women who smoke are less satisfied with their bodies, more concerned about appearance, and have lower self-esteem [50]. The rate of smoking among female college students is related to body image [32,37-39]. These findings suggest that targeting body image concerns may be an effective way to address smoking as part of a multi-behavior intervention. One such study found that a CBT approach, including mindfulness exercises, to address body image produced significantly greater abstinence than a weight management intervention [35].

Concerns About Weight Gain

Studies show that weight concerns play an important role in the smoking behavior of women [34,51]. Compared to nonsmokers, women who smoke are more concerned about becoming overweight [50]. Smoking among women is also related to weight concerns, dieting behavior, and disordered eating symptoms [27,35,52-55]. Concerns about weight also persist after quitting [28]. Weight gain after quitting results from increased dietary caloric intake and decreased metabolic rate [56,57], therefore most behavioral interventions attempt to decrease food intake and/or increase physical activity [6,20,40,42,58,59]. According to a recent meta-analysis of tobacco cessation interventions that aim to prevent weight gain, one of the most effective interventions to increase abstinence and reduce weight gain focused on a cognitive intervention to minimize concern about weight [6].

Self-Efficacy

Self-efficacy refers to the belief or confidence in one's ability to successfully execute a given behavior and is frequently considered one of the central determinants involved in the behavior change process [60,61]. According to self-efficacy theory, individuals' beliefs about their capacity to change a behavior causally influence the outcome when they engage in a behavior change attempt [62]. Self-efficacy theory has been studied widely in the smoking cessation literature. For decades this theory has been used to help identify how confidence in one's ability to quit smoking influences smoking cessation behavior. When people feel confident in their ability to quit smoking, they are more likely to plan to quit [63,64]. This literature suggests that self-efficacy is positively correlated with making plans to quit, especially in the short-term.

Guided Imagery as an Intervention Strategy

Guided imagery is a form of mind-body therapy that involves controlled visualization of specific mental images, and overlaps with mindfulness meditation [65]. Both guided imagery and mindfulness have the core feature of focusing awareness on attention [66]. As shown in Figure 1, guided imagery scripts will target concerns about weight gain, perceptions of body image, and self-efficacy to quit smoking, eat a healthy diet, and engage in physical activity.

It is generally accepted that body image disturbances are associated with disordered eating etiology. Studies have shown that mental imagery is an effective way to address eating disorders, concerns about weight gain, and muscle dysmorphia [67-69]. Meta-analytic reviews have consistently documented the effectiveness of CBT in the treatment of eating disorders [70,71]. Given that mental imagery is an important part of CBT therapy, and this therapeutic modality can effectively address negative body image, we predicted that guided imagery scripts would change women smokers' concerns about body weight and image. Further empirical justification for this prediction is readily gleaned from published studies focused on diet, exercise, and smoking cessation among other health behaviors.

Guided imagery has been shown to improve self-efficacy across a broad range of behaviors, including performance of novel motor tasks [72], prevention of falls with older adults [73], postsurgical functional outcomes [74], and pain management in persons with fibromyalgia [75]. More specific justification for targeting the theorized mediators of smoking cessation using guided imagery is provided in Figure 1.

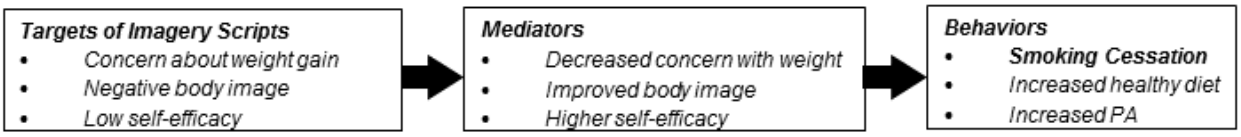
Separate lines of inquiry support the use of mental imagery to enhance physical activity (PA) behavior, improve diet, and smoking cessation [40,58,76-81]. Several recent studies suggest that guided imagery and other mindfulness-based interventions can be effective in assisting smokers to quit [82]. In this study, participants who used an audio CD-based guided imagery intervention were significantly more likely to be abstinent or to have reduced their level of smoking at all follow-up points than control subjects [82]. Another study using imagery resulted in reduced cravings with smokers [80]. Another recent study found that a mindfulness-based intervention attenuated relationships between negative affect and smoking urges, as well as between negative affect and body dissatisfaction [83].

Cross-sectional and intervention studies have clearly documented the impact of guided imagery on exercise behavior [84]. A recent RCT demonstrated that guided imagery scripts effectively increased exercise self-efficacy with sedentary women [78]. The first author (PRG) has conducted numerous studies showing that mental imagery is associated with exercise behavior [85-88]. These studies have shown that individuals who use mental imagery focused on fitness or health goals and exercise technique report greater amounts of exercise than those who do not use imagery. Another randomized trial that used guided imagery among other intervention components resulted in increased exercise behavior after 18-months follow-up as compared to control group participants [89]. In another study, experimental group participants showed increased self-determined exercise behavior after a 10-week imagery intervention as compared to controls [90].

A growing body of literature suggests that guided imagery can be effective at improving dietary behaviors [77,91,92]. One recent study used mental imagery to target implementation intentions in order to increase fruit consumption [91]. Participants randomized to a mental imagery condition targeting implementation intentions demonstrated significantly higher self-reported fruit consumption after 7-days follow-up compared to control, implementation intentions, and goal intentions conditions [91]. These results suggest that mental imagery targeted to specific behavioral intentions may be an effective way to increase goal attainment with dietary behavior. Other studies have also shown that mental imagery is associated with reduced craving or actual consumption of food [77,92,93].

The dual function framework of mental imagery guided the development of the imagery scripts [94]. This framework has recently been supported with psychometric analyses [86]. The dual function framework predicts that mental imagery serves cognitive (eg, skill development) and motivational functions. The guided mental imagery scripts involved images of successfully executing a behavior (eg, walking for exercise) and enhanced self-efficacy, regulation of urges or emotions (eg, anxiety) linked to body image, and goal achievement (eg, losing or maintaining weight).

Figure 1. Project targets, mediators and goal behaviors.



Potential Impact of Mobile Health Apps

A recent study by Spring and colleagues [39] provided evidence to support the use of mobile technology to successfully address multiple behavior changes in diet and physical activity. There is enormous potential reach of mobile apps. As of February 2012, 88% of U.S. adults used a mobile phone, and 53% of these are mobile phones with enabled Internet capacity [95]. In 2012, the top 3 selling mobile platforms in the U.S. were Android (20% of mobile phone owners), iPhone (19%), and Research in Motion (6%) [95]. There are >500,000 mobile software apps (apps) available for the iPhone, Android, and Blackberry devices, which collectively have been downloaded >25 billion times. In 2012, approximately 19% of mobile phone owners had at least one health app on their phone and 52% used their devices to search for health information, and this figure is expected to grow exponentially [95].

Very little research has tapped the great potential of interactive technologies to address multiple risk factors [95]. A mobile phone-based approach offers reach as well as convenience. To our knowledge, the proposed study is the first of its kind to specifically target these underserved smokers using a mobile health platform. If successful, a mobile health app using guided imagery would have broad applicability beyond this intervention and population. The lessons learned in the proposed project could be applied to developing mobile health apps for other chronic conditions with multiple risk factors and the populations at risk for developing those conditions.

The systematic development and evaluation of a mobile health app is novel. According to Abroms and colleagues [96], the currently available mobile apps for tobacco cessation are not evidence based and do not provide effective treatment. Backinger and Augustson [97] called for the systematic development of mobile apps in this area to provide efficacious treatment for tobacco cessation. Our proposed study would be the first of its kind to create and test a mobile app to address smoking, diet and physical activity in an adult population.

Successful studies of multiple risk factor interventions have traditionally been conducted in in-person, individual, or group settings [97]. A growing mismatch between the demand for health-related services and supply has burdened our health care system [98-103]. The use of mobile technologies can help to reduce some of this burden by replacing a proportion of face-to-face encounters with a model that emphasizes patient self-management, independent of time and place. Mobile technologies allow users 24/7 access to information and resources, as well as simple tools for self-monitoring, an important component of any health behavior change intervention. Mobile apps are also highly customizable, and can be easily configured by the individual patient to provide tailored feedback in support of behavior change. These features increase the likelihood that critical information and feedback is delivered to the patient when they need it most. Further, the flexibility of mobile apps allow capture of individual patterns of behavior (eg, cravings); data which can then be correlated with diet and physical activity behaviors (and other data) to better understand the factors related to smoking.

Methods

The proposed project produced an interactive mobile phone app to deliver a guided imagery intervention targeting concerns about body weight, body image, and self-efficacy in order to increase tobacco cessation, healthy eating, and physical activity. We completed the following: Aim 1) created program content and designed the functionality of the prototype app for use on the Android platform, in collaboration with 9 participants in multiple focus groups (several participants attended more than 1 focus group) and 1 in-depth interview; Aim 2) programmed and tested the app's usability with 6 participants; and Aim 3) we are currently pilot testing the feasibility and acceptability of the app, evaluating the relationship of program use to tobacco cessation, dietary behaviors, and physical activity, and assessing consumer satisfaction with 73 weight-concerned women smokers who own an Android-based mobile phone. In a future R01, we will test the efficacy of the app in a randomized controlled trial.

Project Time Line

This project occurred in several stages over 2 years. Development of the content and user interface occurred over the first 6 months of Year 1. Based on the results of focus group testing, the prototype mobile app was programmed, user testing and program refinement then occurred over the last half of Year 1. The feasibility trial is currently taking place (Year 2), and includes programming of the informed consent and study data collection tools, participant recruitment, data collection, data analysis, report writing, and development of the Manual of Procedures for the future randomized trial.

Program Development Process

During Stage 1, the team collected content materials, reviewed other health-related cell-phone apps, and outlined the desired interface design, functionality, and content for the prototype app. Then, the team drafted content elements for the intervention (see Textbox 1). Five guided imagery audio files were created (1 introductory file, 3 behavior-specific files, and 1 general file) and 3 resource pages related to smoking cessation, diet, and physical activity were developed. These resources pages addressed the common barriers experienced by women smokers when quitting (eg, cessation resources, weight-loss resources, physical activity resources). The app provided suggestions for addressing these barriers and a direct link to each resource, based on user input. We also provided a daily diary/tracking calendar to track use of the guided imagery scripts, tobacco use/cessation variables (eg, cravings, withdrawal symptoms and number of cigarettes smoked), diet variables (eg, increased fruit/vegetable intake), and physical activity variables (eg, frequency, intensity and type of exercise). These user inputs were designed to tailor messages around the benefits of continued practice with the scripts and provide suggestions for alternative strategies (eg, behavioral support for cessation, including a "one touch" call to their local tobacco quitline). The technology was developed to promote user input and interactivity, and time spent listening to mental imagery files. Many program elements, such as the tracking calendar and data collection and feedback, were refined based on responses from focus group participants.

Textbox 1. App components.

Guided imagery audio files

1. Introduction to Guided Imagery
2. Be Smoke-Free
3. Eat Well
4. Get Moving
5. Feel Fantastic

Resources for tobacco cessation

1. Direct connect to tobacco quitlines
2. Links to evidence-based information

Daily diary

1. Smoking
2. Eating 5 fruits and vegetables
3. Getting 30 minutes of exercise

Tailored feedback based on user input

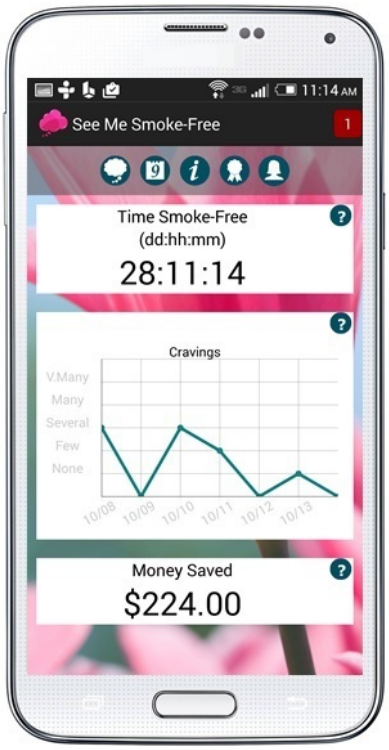
1. Awards for meeting goals

Motivational messages

In Stage 2, the programming team prepared functional specifications for the software platform and designed the interactive features. These features included the User Profile function that facilitates participants' tailoring of the program (eg, set a quit date, set time for delivery of guided imagery files). In addition, the entire team designed the overall infrastructure to accommodate user registration, baseline surveys, send/receive functions and within-app navigation. All design features were incorporated into a graphic storyboard that described the prototype and illustrated the app's important functions, then presented to focus group participants for their review. Participants helped refine each of the proposed program components or suggest additions. Participants provided feedback on length of imagery audio files, narrator voice, resources and information, elements of the user interface, content and timing of messages. In Stage 3, based on feedback from focus groups, we revised some of the program components while in stage 4, program assets elements (eg, text, graphics, images, resource materials) were finalized and programmed into the app.

Stage 5 focused on development of the "Alpha" version of the app. Programmers created rapid prototypes of the app that allowed the project team to review and confirm that app functionality, content and design were consistent with the overall goals of the program. After internal testing of the Alpha version, we recruited groups of participants for usability testing, which yielded a final set of changes prior to feasibility testing. The app was fully automated and programmed using the Android application programming interface (API). The app takes full advantage of all of the functionality made available by the Android API including push notifications, saving and retrieval of local data, sending/retrieval of data over the network, and interfacing with the project database. The tight integration of data and programming allowed us to tailor app content (eg, deliver content based on quit date or program use) as well as unobtrusively track all usage details and user choices. The developers built the app to accommodate expansion of the content and the migration of the app to other mobile platforms in future projects (Figure 2).

Figure 2. See Me Smoke Free app home screen.



Evaluation Overview

We used qualitative and quantitative methods and conducted focus group and usability testing with 9 participants in 4 focus groups and 7 in-depth interviews (several participants attended more than 1 focus group or interview), as well as feasibility and consumer satisfaction testing of the prototype product with 73 participants.

Participant Recruitment and Eligibility

Participants were female cigarette smokers who were interested in quitting. Additional eligibility requirements include having smoked for ≥ 1 year, being at least 18 years old, having Internet and e-mail access, and speaking English.

Local participants were recruited via University social media and other websites, Craigslist, recruitment flyers in health care clinics and other community locations, and news coverage in local media outlets. Potential participants for focus and user groups were contacted via phone by trained project staff who described the research study and invited individuals to participate. Participants recruited for focus groups and user groups resided in the greater Tucson, Arizona area. Focus group participants were considered eligible if they owned any type of mobile phone.

Participants recruited for the feasibility test could be located anywhere in the United States. Local and national press coverage about the study and no-cost Facebook, Twitter, and other social media sites helped spread the word about recruitment for the study. Informed consent and all data collection were conducted via the mobile app. After a participant was consented, project staff collected contact information, including the participant's cell phone number, email, and mailing address. Usability test and feasibility test participants needed to own an Android phone to be eligible to participate. All participants received a US $50 check for completing the study.

Formative and Process Evaluation

Focus Groups

A total of 9 women smokers (7 White, 1 Hispanic/Latino, and 1 White/Native American) participated in four 1.5-hour focus groups and 1 in-depth interview to offer input about the prototype design. During the focus groups, participants were asked to discuss topics that directly informed the refinement of our app with respect to program presentation, features (eg, daily diary), script content, and potential tailoring of content based on differences in body image [104-106]. We followed recommended guidelines for focus groups [107,108]. Session transcripts were analyzed for salient constructs, issues, and language use to inform further program development. The first and second focus groups reviewed the 5 imagery scripts developed by the team. After the first 2 focus groups, the imagery scripts were refined and the prototype design modified for presentation to the third and fourth groups for input.

Usability Testing

A total of 6 participants were recruited to test user interactions with the Alpha prototype. Testing usability with 5 users per population of interest is generally deemed sufficient based on established guidelines [109]. As participants used the Alpha prototype, they were asked to think aloud, verbalize their reactions as they viewed screens, and describe their navigational choices as they made them. Participants performed each of the major tasks the app supports. Interviewers took detailed notes so that comments could be matched to screens or technical functions that elicited feedback. Participant responses were characterized as positive or negative reactions to the features and functions, and difficulty or facility with using the app or

interface. Usability testers also completed the 10-item adaptation of Brooke's widely-used and validated System Usability Scale and a consumer satisfaction questionnaire used in the principal investigator's (JG) previous research [109]. After each tester provided feedback, revisions were made to the Alpha prototype, and additional user tests were conducted. Usability testing took approximately 1½ hours per session.

Feasibility and Satisfaction Testing

The feasibility and acceptability of the final Alpha prototype is being evaluated in an open, pre/post trial with 73 women smokers with Android mobile phones.

Study Procedures

Participants downloaded the app from the Google Play store. After launching the app, participants were asked if they would like to participate in research. If so, they were screened for eligibility, and eligible respondents provided informed consent and completed baseline assessments. Project staff were available to assist if a participant experienced difficulty, and any technical problems encountered by participants were documented. Participants were asked to use the app "most days" for 30-days. During this period, participant use and engagement data were collected. Prompts for the follow-up assessments were delivered and completed via the mobile phone or via a weblink.

Data Collection

Demographic and Descriptive Variables

Demographic information collected at baseline included year of birth, ethnicity, and socio-economic status (SES).

Tobacco Use History, Current Use Patterns, and Nicotine Dependence

Use of all tobacco products were assessed using a series of questions that have been standardized and employed in previous studies [110-112], and the Fagerström Tolerance Nicotine Dependence Scale [113].

Body Mass Index

Self-reported height and weight will be converted to body mass index (kg/m^2).

Concern about Weight Gain

We used items from the Weight Control Smoking Scale [114] and the Concern about Post-Cessation Weight Gain Scale [115] to measure concerns about weight, use of smoking to control weight, and concerns about gaining weight as a result of quitting smoking. Both measures have been used extensively and have demonstrated reliability and validity [114].

Self-Efficacy

We assessed the degree of confidence to prevent weight gain after quitting smoking with the *Weight Efficacy after Quitting Scale*, and self-efficacy for quitting with the 15-item version of the Condiotte & Lichtenstein Confidence Questionnaire [116].

Body Image

The Multidimensional Body-Self Relations Questionnaire [117] provides a standardized, attitudinal assessment of body image. We chose the 7-item Appearance Evaluation Subscale [117,118] which measures feelings of physical attractiveness and satisfaction or dissatisfaction with one's appearance [119,120].

Diet

To assess the daily number of fruit servings and vegetable servings, the 2-item interactive version of the National Cancer Institute Fruit and Vegetable Scan was used [121]. Also used were several questions from the Dietary Screener Questionnaire used in the National Health and Nutrition Examination Survey (NHANES).

Physical Activity

We assessed physical activity with the Godin Leisure-Time Exercise Questionnaire (LTEQ) [122]. The LTEQ has a long history of use [123] and is simple to administer using a mobile app. It is also sensitive to mild, moderate, and strenuous exercise and is scored in two ways: total number of minutes of activity or a metabolic equivalents (METS) expenditure estimate.

Tobacco Use Outcome Measures

As recommended by the Society for Research on Nicotine and Tobacco [124] and Velicer and Prochaska [125] we used several self-report measures, including prolonged abstinence and point prevalence.

Withdrawal and Cravings Symptoms

All participants were asked to rate their experience with withdrawal symptoms and cravings using a 5-point rating scale of severity 3 times per day at random intervals [126]. To do so more often would have created a confounding effect on the intervention, and may also have increased response burden to unacceptable levels.

App Satisfaction and Acceptability

At follow-up, we will use Tullis and Stetson's 8-item adaptation [127] of Brooke's widely used System Usability Scale to rate the usability of our app [109]. We also asked participants about their satisfaction with the program, perceived usefulness of information, relevance to smoking cessation efforts, whether they would recommend the program to others.

App Use

We monitored (1) the length of time participants interact with the app, (2) the number of screens they visit within the app, (3) use of the daily diary, and (4) the number of links they click on while using the app.

Data Analysis

Focus Group and Usability Testing

Interviews conducted during focus groups were audio-recorded, and cross-referenced with notes taken by trained staff. User testing interviews were conducted by 2 trained staff, with one in charge of interacting with the participant and the other responsible for detailed note-taking. Transcripts and notes were analyzed by several members of the project team with extensive expertise in analyzing focus group and user group data for common themes, changes to the program content and/or functionality, and suggestions for improvement.

Feasibility and Acceptability Testing

The feasibility and acceptability of the program is being evaluated in an open trial with 73 participants. A pre/post design will evaluate participants' reported usability and satisfaction with the product, reported abstinence from smoking, and actual use of the program. While this design does not control for potential threats to internal validity, it will allow for the initial evaluation of the program's impact on cessation. Threats to internal validity will be addressed in a subsequent RCT.

The feasibility of the program will be demonstrated by the achievement of the following benchmarks: (a) recruiting at least 50 women to use the app regularly within a 3-month window, (b) a high level of app usage by the participants (eg, 80% of participants using the program regularly during the data collection phase), and (c) based on our previous experiences of program usage, a retention rate of at least 70% at the 3-month assessment. Acceptability will be defined as a high degree of consumer satisfaction (eg, mean usability and satisfaction ratings >4 on a 5-point scale).

Tobacco Use, Diet, and Physical Activity

We will assess the absolute cessation rate of participants, the smoking rate for nonquitters and the number of quit attempts. Paired *t*-tests will be used to evaluate change in each of these cigarette consumption measures from baseline to 30 days post, and again from baseline to 90 days post. With 50 participants, significance level = 0.05, there is 0.80 power to detect change of d=0.66, a medium effect size. We will also use multilevel modeling for repeated measures to examine change in each food/beverage consumption measure across all 3 waves (baseline, 30 days and 90 days), with level-1 units consisting of the repeated measures for each participant, and the level-2 unit being the participant. (Note that multilevel modeling is tolerant of missing waves of data.) For each wave, we will also construct composite measures of concern about weight gain (a composite of 3 Likert-type items), perceived self-efficacy regarding smoking cessation (5 Likert-type items), body image (7 Likert-type items), mental imagery (4 Likert-type items), and diet (4 items assessing juice, lettuce, vegetable and water consumption). Body weight/BMI and the frequency and intensity of exercise will be measured as well. Multilevel modeling will be used to examine changes in these measures across the 3 waves. Finally, multilevel modeling will also be used to examine whether weight gain, perceived self-efficacy regarding smoking cessation, body image, diet, body weight/BMI and exercise have an association with the absolute cessation rate of participants, the smoking rate for nonquitters and the number of quit attempts.

Program Use and Engagement

The See Me Smoke-Free (SMSF) app allows monitoring of all app usage by study participants through the intervention database. However, we will also use Google Analytics which allows us to track a limited number of app functions for both participants and nonparticipants. Participants will have all app usage tracked in the intervention database with regular synchronizations from the SMSF app. The analytics provided by Google include anonymized information for the users, differentiating by a textual tag if the user is a participant or nonparticipant. It tracks the following actions: visit the app home screen; set or update a quit date; start or finish listening to an audio file. Thus such statistics are available daily for study participants and nonparticipants. We will examine individual metrics (ie, visit duration, number of type of screens viewed, change in usage patterns over time), and interactive activities (eg, answer daily questions) engaged in for study participants. We will also aggregate these data into composite exposure scores that combine number of visits and overall visit duration to calculate correlations between outcome and measures of program exposure and engagement [128,129]. Finally, we will compare app use on visits to the home screen, setting or updating a quit date, and listening to the audio files between participants and nonparticipants using the data from Google analytics.

Results

The study was started January 1, 2014. The app was launched and feasibility testing began in April 1, 2015. Participants were enrolled from April 1-June 30, 2015. During that time, the app was downloaded over 350 times using no paid advertising. Participants were required to use the app "most days" for 30 days or they would be dropped from the study. We enrolled 151 participants. Of those, 78 were dropped or withdrew from the study, leaving 73 participants. We have completed the 30-day assessment, with a 92% response rate. The 90-day assessment is ongoing. During the final phase of the study, we will be conducting data analyses and disseminating study findings via presentations and publications.

Discussion

Preliminary Results

This paper presents the protocol used in an ongoing study to develop and test a guided imagery intervention targeting smoking, diet and physical activity among women. The multi-behavioral intervention is delivered via an mHealth app, called See Me Smoke-Free. We used an iterative approach to development, including focus groups, individual interviews, and user testing, to develop the app content, functionality, and user interface. The app was successfully deployed to the Google Play store, and downloaded hundreds of times. We learned many lessons about the challenges faced when developing technology to be delivered using the Android platform, recruiting participants with no in-person contact, and gathering data from multiple sources.

Future Directions

We are currently preparing 2 manuscripts that describe in detail our recruitment and retention strategies and our development process. We are in the final stages of data collection and have begun data analyses. The results of these analyses will be described in a future outcome paper.

Acknowledgments

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Conflicts of Interest

None declared.

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Abbreviations

API: application programming interface
BMI: body mass index
CBT: cognitive behavioral theory
COPD: chronic obstructive pulmonary disease
LTEQ: Godin Leisure-Time Exercise Questionnaire
METS: metabolic equivalents
NHANES: National Health and Nutrition Examination Survey
PA: physical activity
RCT: randomized controlled trial
SMSF: See Me Smoke-Free

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Protocol

Development of a Decision Aid for Cardiopulmonary Resuscitation Involving Intensive Care Unit Patients' and Health Professionals' Participation Using User-Centered Design and a Wiki Platform for Rapid Prototyping: A Research Protocol

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Abstract

Background: Cardiopulmonary resuscitation (CPR) is an intervention used in cases of cardiac arrest to revive patients whose heart has stopped. Because cardiac arrest can have potentially devastating outcomes such as severe neurological deficits even if CPR is performed, patients must be involved in determining in advance if they want CPR in the case of an unexpected arrest. Shared decision making (SDM) facilitates discussions about goals of care regarding CPR in intensive care units (ICUs). Patient decision aids (DAs) are proven to support the implementation of SDM. Many patient DAs about CPR exist, but they are not universally implemented in ICUs in part due to lack of context and cultural adaptation. Adaptation to local context is an important phase of implementing any type of knowledge tool such as patient DAs. User-centered design supported by a wiki platform to perform rapid prototyping has previously been successful in creating knowledge tools adapted to the needs of patients and health professionals (eg, asthma action plans). This project aims to explore how user-centered design and a wiki platform can support the adaptation of an existing DA for CPR to the local context.

Objective: The primary objective is to use an existing DA about CPR to create a wiki-based DA that is adapted to the context of a single ICU and tailorable to individual patient's risk factors while employing user-centered design. The secondary objective is to document the use of a wiki platform for the adaptation of patient DAs.

Methods: This study will be conducted in a mixed surgical and medical ICU at Hôtel-Dieu de Lévis, Quebec, Canada. We plan to involve all 5 intensivists and recruit at least 20 alert and oriented patients admitted to the ICU and their family members if available. In the first phase of this study, we will observe 3 weeks of daily interactions between patients, families, intensivists, and other allied health professionals. We will specifically observe 5 dyads of attending intensivists and alert and oriented patients discussing goals of care concerning CPR to understand how a patient DA could support this decision. We will also conduct individual interviews with the 5 intensivists to identify their needs concerning the implementation of a DA. In the second phase

of the study, we will build a first prototype based on the needs identified in Phase I. We will start by translating an existing DA entitled "Cardiopulmonary resuscitation: a decision aid for patients and their families." We will then adapt this tool to the needs we identified in Phase I and archive this first prototype in a wiki. Building on the wiki's programming architecture, we intend to integrate the Good Outcome Following Attempted Resuscitation risk calculator into our DA to determine personal risks and benefits of CPR for each patient. We will then present the first prototype to 5 new patient-intensivist dyads. Feedback about content and visual presentation will be collected from the intensivists through short interviews while longer interviews will be conducted with patients and their family members to inform the visual design and content of the next prototype. After each rapid prototyping cycle, 2 researchers will perform qualitative content analysis of data collected through interviews and direct observations. We will attempt to solve all content and visual design issues identified before moving to the next round of prototyping. In all, we will conduct 3 prototyping cycles with a total of 15 patient-intensivist dyads.

Results: We expect to develop a multimedia wiki-based DA to support goals of care discussions about CPR adapted to the local needs of patients, their family members, and intensivists and tailorable to individual patient risk factors. The final version of the DA as well as the development process will be housed in an open-access wiki and free to be adapted and used in other contexts.

Conclusions: This study will shed new light on the development of DAs adapted to local context and tailorable to individual patient risk factors employing user-centered design and a wiki to support rapid prototyping of content and visual design issues.

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KEYWORDS

cardiopulmonary resuscitation; end-of-life planning; goals of care discussions; intensive care medicine; medical informatics; shared decision making; user-centered design; wikis

Introduction

Background

Over the last 90 years, Canadians have gained an average of 24.6 years of life expectancy according to the Conference Board of Canada [1]. Over the same period, the most common place of death has shifted from home to hospital [2]. Therefore, a growing number of older and sicker patients are being admitted to an intensive care unit (ICU) where either they or their family members will have to make crucial decisions about their goals of care [3]. One of these decisions is to decide whether or not to conduct cardiopulmonary resuscitation (CPR) in the event of a cardiac arrest.

Data from the Get with the Guidelines-Resuscitation registry, the world's largest database on in-hospital cardiopulmonary arrest from over 400 participating hospitals in the United States, estimate that the incidence of treated cardiac arrest is 0.92/1000 patient days. Almost half of these events (48%) occur in the ICU [4]. Over the years, CPR has become a standard intervention used in almost all cases of cardiac or respiratory arrest unless a do-not-resuscitate order (DNR) is recorded in the patient's chart [5]. However, estimated survival to discharge after treatment for in-hospital cardiac arrest is 18%, with about half of these patients being neurologically intact or having only mild neurologic deficits at discharge [4].

Because cardiac arrest can have potentially devastating outcomes even if CPR is performed, making the decision about whether or not to initiate CPR should involve the patient or his/her surrogate decision maker(s) in cases where the patient is unable to participate in decision making [4]. Discussions about advance directives for CPR in the ICU are a major source of stress and grief for patients, their family members, and their health care professionals. Shared decision making (SDM), a process in which health care professionals and patients work together to make health care choices, offers opportunities to facilitate discussions about advance directives regarding CPR in ICU settings [6].

A common way to support SDM is the use of patient decision aids (DAs). DAs are generally used in situations where a range of treatments are possible, with each treatment having its own advantages and disadvantages. They are intended to be used by patients to complement health care professionals' counselling. DAs aim to clearly present the best available evidence and to support patients' reflections about their values and preferences [7].

There exists a series of multiplatform DAs about CPR developed and owned by ACP Decision, an American nonprofit foundation, and adapted to various care settings, illness, and decision makers (patients or surrogate decision makers). Although these DAs are available in multiple languages, there currently lacks a French version that could be used by the population in the Province of Québec where the predominant language is French. Moreover, ACP Decision's DAs are currently only available to organizations within the United States. We were granted permission to adapt another DA about CPR that was developed in Canada, the "Cardiopulmonary resuscitation: decision aid for patients and their families" [5]. However, in addition to being only available in English, it was initially designed for patients hospitalized in the general hospital ward. Patients admitted to the ICU are critically ill and have specific contextual factors (eg, urgency to make a decision, difficulty reading, unstable condition) that need to be considered in the creation of a DA designed for their use. Moreover, this DA only presents population-level statistics about the outcomes of CPR classified by age category and by the reason of cardiac arrest. It does not present a precise and tailored survival estimate for each patient. The Good Outcome Following Attempted Resuscitation (GO FAR) clinical prediction rule [4] has recently been developed to help health care providers produce better estimates of patients' likelihood of survival after an episode of in-hospital cardiac arrest, with these patients being neurologically intact or having

only minimal deficits at discharge (Cerebral Performance Category 1 [CPC 1]). The GO FAR clinical prediction rule assigns scores to 11 risk factors such as major trauma, renal insufficiency, and septicemia among others and 1 protection factor (ie, being neurologically intact before cardiac arrest). Using these variables, a final estimate of the survival rate with minimal neurological deficits after in-hospital CPR is generated. The final survival-to-discharge rates with CPC 1 range from a maximum of 27.7% to a minimum of 0.9% [4].

Adapting knowledge tools such as DAs to local context is a crucial step in knowledge translation because it (1) reduces duplication of effort and optimizes use of existing resources; (2) encourages consideration of implementation and "fit" with the local context; (3) enhances applicability; and (4) engages local knowledge users, thus increasing the likelihood of uptake [8]. This step therefore requires engaging local knowledge users to ensure that the knowledge is relevant and applicable to their needs, from its generation to its implementation [9]. Although this is a key step in knowledge translation, little is known about how to accomplish this step effectively [10-12]. Experts have referred to this problem as an "evidence-based crisis" [10,13] and have challenged researchers to find innovative solutions that support the involvement of local knowledge users in adapting knowledge tools to their contexts [14-17].

We submit that using a wiki, a website that can be consulted and edited by anyone who is granted access, could be an effective strategy for adapting DAs to local contexts, as wikis were precisely designed to involve users interactively in the generation and application of knowledge [18]. Wikis—highly accessible, interactive communication vehicles—have also been shown to increase professionals' self-efficacy with regard to their use of various types of knowledge tools [18-20]. The use of a wiki to perform rapid prototyping has successfully helped in the creation of knowledge tools adapted to the needs of patients and health care professionals [18,21-26]; however, to the best of our knowledge, our team is the first to use a wiki to support the creation of a patient DA.

Objectives

The primary objective is to use an existing DA about CPR to create a wiki-based DA that is adapted to the context of a single ICU and tailorable to individual patient risk factors while employing a user-centered design. The secondary objective is to document the use of a wiki platform for the adaptation of knowledge tools such as patient DAs.

Methods

We will employ a user-centered design and a rapid prototyping method to involve patients and other end users in the development process. User-centered design is a longstanding and proven framework and methodology for the development of products, services, and systems [8,27-29]. User-centered design is a highly iterative method for optimizing the user experience—and thus the effectiveness—of a system, service, or product [27]. In this framework, a user is any person who interacts with (in other words, "uses") the system, service, or product for some purpose.

Phase I: Ethnographic Needs Assessment

The main researcher (AP) will conduct 3 weeks of ethnographic observation of patients, families, intensivists, and other allied health professionals' daily interactions. Ethnography is a qualitative method involving the immersion of the researcher into the setting to be studied. Traditionally used in cultural anthropology, ethnography is increasingly being used in health care research. Ethnography allows discovering rich information that could not have been discovered through surveys or other quantitative methods [30]. Because the presence of the main researcher in the ICU will not interfere with the usual care process and that the main goal of these observations is to learn how to improve the decision-making process in the ICU, only verbal consent will be required by the Ethics Review Committee to complete these observations.

During the observation sessions, the main researcher will specifically observe 5 dyads of attending intensivists and alert and oriented ICU-admitted patients, and their family members if available, who need to clarify whether or not to initiate CPR in case of a cardiac arrest. Patients who are unstable, aged less than 18 years, have cognitive impairments, and who do not speak French will be excluded. Potential participants will be identified by attending intensivists. Aptitude of the patients to participate will be confirmed at the beginning of the observation by their attending intensivist using a short evaluation of their orientation in time, space, and person. Using an observation grid developed by a human factors engineer (HW), the main researcher will observe the discussion and note users' needs, goals, strengths, and limitations of the actual process of decision making without the use of a DA. The first 5 eligible patients will be enrolled in the study. The following data will be collected from the patient's chart: date of birth, sex, reason of ICU admission, and level of medical intervention requested on admission to the ICU (full code, limited intervention, DNR, patient undecided or information not available).

In addition to ethnography, the main researcher will conduct one 60-minute semistructured interview with each of the 5 intensivists involved in the project. The development of the interview grid will be informed by the data collected during the ethnography. We will also collect the following demographic information about the 5 intensivists: age, sex, clinical specialty, and number of years working in the ICU.

Phase II: Rapid Prototyping

Development of the First Prototype

The needs gathered in the first phase will inform the construction of the first CPR DA prototype that will be built according to the International Patient Decision Aid Standards [31]. To develop this first prototype, we will start by translating and adapting the existing "Cardiopulmonary resuscitation: a decision aid for patients and their families" [5] using a recognized method for cultural adaptation and translation [32]. This method will require that 2 translators produce 2 translations from English to French. These 2 versions will then be merged and backtranslated to English and presented to the original authors to identify any discrepancies that need to be addressed. We will also translate the GO FAR score [4] into French using the same

method. Using the programming architecture of a wiki platform, we aim to integrate the GO FAR rule into our DA so that patients can personalize the calculation of their own survival rate after CPR. The 5 intensivists and other allied health professionals working in the ICU at Hôtel-Dieu de Lévis, Québec, Canada will be granted access to the wiki platform and will be requested to report any content and visual presentation issues using the collaborative writing functionality of the wiki. Any major issues will be addressed prior to the beginning of the rapid prototyping with actual patients and family members.

Rapid Prototyping Cycles

Rapid prototyping will involve 15 new patients admitted to the ICU and their family members if available. Alert and oriented patients needing to discuss their goals of care or needing to validate goals of care previously discussed will be identified by the main researcher in collaboration with the attending intensivist. A sample of 5 eligible patients will be enrolled at each of the 3 rapid prototyping cycles. Previous uses of this method suggest that a sample of at least 15 participants is adequate to detect over 90% of all usability problems related to products [33].

Inclusion and Exclusion Criteria for Phase II

Patients will be alert, oriented, and capable to consent to participate in our project (inclusion criteria). This will be determined by the attending intensivist. The exclusion criteria are as follows: patients who participated in the first phase, with cognitive impairments and/or critically unstable conditions, aged less than 18 years, and those who do not speak French. Family members of the participating patients will also be invited to participate if available at the time of the study, but this will be optional.

Consent to Participate and Recruitment

Potential participants will be identified by the main researcher who will first obtain consent from the attending intensivist to approach the patient. Although we will purposively attempt to target a wide range of different types of patients based on their age, sex, and disease process, these participants will form a convenience sample based on their availability at the time of recruitment. Once the intensivist judges that the eligible patient is capable and fit to participate, the main researcher will obtain written consent from the patient to participate and will then present the DA to the patient and/or a family member. A minimum of 3 hours will be given to the participant to read the document. A follow-up meeting will be arranged 3 hours later with the attending intensivist so that the patient can provide his/her feedback on our DA and engage in a discussion about their goals of care.

Intervention and Data Collection

The patient/family member and the intensivist will be requested to review together the clinical content and visual aspect of the wiki-based DA. Using an observation grid developed by a human factors engineer (HW), the main researcher will observe users' needs regarding our DA and will assess its strengths and limitations. Then, the main researcher will conduct interviews with the patients and/or their family members. The interview will start with 3 sociodemographic questions about the participants' highest level of education completed, their profession, and their religion. Then, 7 open-ended questions will follow about the DA concerning the following:

- Clarity of information
- Social acceptability of the information presented
- Relevance of the information presented
- Preferred element of the DA
- Suggested improvements to be made to the DA

Finally, each participant will be asked if they had previously discussed their goals of care and their preferences about CPR. We will also verify if our DA changed their previous advance directives (eg, changing from wanting to receive CPR to now refusing CPR). After each use of our DA with any patient, intensivists will also be requested to provide feedback about their experience using the DA with their patient. All interviews will be recorded and then transcribed verbatim.

We will also collect the following data from the patient's chart: date of birth, sex, reason for ICU admission, and the code status preferences upon admission to the ICU.

Data Analysis

After each rapid prototyping cycle, 2 researchers (AP and PA) will perform qualitative content analysis of the verbatim transcripts, audio recordings, and researcher interview notes and observations to identify the usability problems (eg, visual design, format, layout of information), and need for content clarification. Any problem identified will be addressed prior to the next round of user testing. All changes to the prototype will be done online using the wiki platform to keep track of the changes and the different prototype versions produced.

We will perform descriptive statistical analyses of the patient and intensivist sociodemographic data. We will also measure the proportion of patients who state that they changed their goals of care after reading our DA and will note if this change increased the level of care or decreased it. We will also compare patient's final decision about CPR after reading our DA with the level of care documented in the medical chart.

Ethical Considerations

This study was approved by the Research Ethics Board of the Centre Intégré de Santé et de Services Sociaux de Chaudière-Appalaches on January 28, 2015 (CER-1415-019). Informed written consent will be obtained from patients and family members participating in Phase II. Participants will have the opportunity to withdraw from the study at any time. Quantitative data gathered about the participants and qualitative information gathered during the observation sessions and interviews will be kept strictly confidential and the study results will not allow to identify participants. Five years after the publication of the study results, all information about participants will be destroyed. If the participant or a family member of the participant demonstrates by his or her words, gestures, or behavior any kind of discomfort with respect to information contained in the decision tool, the situation will be reported to the attending intensivist. Patients will not be paid for their participation. Because family members participating in the prototyping phase could be asked to travel to the ICU to

meet our research team, they will be offered a small stipend to pay for parking or a meal at the hospital's cafeteria.

Results

We expect to create a wiki-based DA about CPR adapted to the local needs of patients, their family members, and their attending intensivists in a single ICU and tailorable to each patient's characteristics. The final content and visual aspect of the DA will be influenced by the comments received and interactions observed with the 15 dyads in the rapid prototyping phase. This study will be conducted between July and December 2015. We expect that the final version of our DA as well as a detailed description of the steps used to develop it will be available online at the beginning of 2016.

Discussion

Our study proposes a quick, efficient, and low-cost development methodology for DAs adapted to patients' needs and to local care settings. As the population is aging and health care costs are increasing, health systems need a large number of DAs based on constantly evolving evidence. This is even more pertinent in the Province of Quebec with the implementation of and end-of-life law that regulates end-of-life care and advance directives planning [34]. Aside from the more controversial aspects of this law (ie, physician-assisted dying) this law mandates the creation of a province-wide register of patient's advance directives. This register will contain advance directives prepared by patients given by a notarial act or in the presence of a witness on a form approved by the Minister of Health. The creation of this register will increase the need for highly usable and accessible DAs that can support patients' decision making about end-of-life questions such as the one about CPR. We expect that using a wiki will help health professionals and researchers across Quebec, Canada, and elsewhere adapt our DA to their own context and culture, thus reducing duplication and accelerating the dissemination of such DAs for the benefit of more patients.

Thus, this study will enrich the emerging literature on the use of wikis and other collaborative writing applications to create and disseminate knowledge application tools, as well as applying user-centered design to develop DAs that better support SDM.

Limitations

Our study protocol has some limitations. First, although studies on usability testing suggest that 15 patients are sufficient to address most usability issues, we cannot ensure that our final DA will address all the issues about end-of-life decisions that are very complex, emotionally charged, and culturally sensitive. Our wiki platform will however offer an interesting solution to facilitate making improvements to our local DA if new usability or content issues arise in the future. Second, the vast majority of patients admitted to our ICU setting will be white, Catholic, and French-speaking while the issues of end-of-life decisions are deeply linked to culture and religion [35]. This will limit the broader use of our context-adapted DA to settings that are more multicultural. Nevertheless, the use of our wiki platform and methodology will still help other researchers and health professionals to adapt our open-source and free tool to these other cultural settings. Third, although we aim to recruit as many family members as possible to make our DA fully adapted to their needs as well, their participation will be optional for feasibility and ethical reasons. Future studies will have to explore the development of DAs adapted to the needs of surrogate decision makers in situations where patients are unable to make decisions for themselves. Finally, this study will not measure the impact of creating a context-adapted DA. A prospective study assessing the impact of our DA on patient satisfaction, quality of care, and patients' decisional conflict will be needed.

Conclusion

This study will enrich the emerging literature on the use of wikis and other collaborative writing applications to adapt knowledge tools to the local context, as well as applying user-centered design to develop DAs that better support SDM. We expect that using a wiki will help other centers and researchers adapt our DA to their own context and culture, thus reducing duplication and accelerating the dissemination of such DAs.

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Conflicts of Interest

None declared.

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Abbreviations

CPC 1: Cerebral Performance Category 1
CPR: cardiopulmonary resuscitation
DA: decision aid
DNR: do-not-resuscitate order
GO FAR: Good Outcome Following Attempted Resuscitation
ICU: intensive care unit
SDM: shared decision making

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Original Paper

A Blended Intervention for Patients With Knee and Hip Osteoarthritis in the Physical Therapy Practice: Development and a Pilot Study

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Abstract

Background: Blended care, a combination of online and face-to-face care, is seen as a promising treatment option. However, actual use of blended interventions in practice is disappointing.

Objective: The objective of this study was two folded. The first aim was to develop a blended exercise therapy intervention for patients with knee and hip osteoarthritis that matches the values of the users and that can be implemented in the daily routine of physical therapists. The second aim was to investigate the feasibility through interviews and a pilot study.

Methods: In this paper, we employed the first 3 steps of the CeHRes road map to develop a blended intervention for patients with knee and hip osteoarthritis. We used interviews, a focus group and discussions with stakeholders to explore the needs, values, and requirements with respect to our to-be-developed blended intervention, which we called e-Exercise. The first version of e-Exercise was tested in a pilot study. Feasibility outcomes, including recruitment rates within each practice, website usage (assignments completed and website visits), and user satisfaction, were measured. In addition, therapists and patients from the pilot study were interviewed to investigate users' experiences.

Results: The study captured important information about stakeholders' needs and perspectives. Based on our findings, we created a first version and attuned the application's content, functionality, and structure. Patients and, to lesser extent, physical therapists were satisfied with the e-Exercise intervention. Eight patients were recruited by 8 physical therapists. Of the 8 patients, 6 completed more than 7 of 12 modules.

Conclusions: This study outlines the development and feasibility of a blended exercise therapy intervention for patients with knee and hip osteoarthritis. E-Exercise offers an alternative approach in the physical therapy treatment of knee and hip osteoarthritis. This study provides valuable information to conduct a further trial to evaluate the (cost) effectiveness of e-Exercise compared to usual physical therapy.

Trial Registration: Netherlands Trial Register Number: NTR4224; www.trialregister.nl/trialreg/admin/rctview.asp?TC=4224 (Archived by WebCite at http://www.webcitation.org/6fOK4lrTO).

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KEYWORDS

development; pilot study; osteoarthritis; blended care; eHealth; physical activity

Introduction

Knee and hip osteoarthritis (OA) are leading causes of disability in older people [1]. In the upcoming years, the number of people with knee and hip OA will grow due to the aging population and escalating risk factors, such as obesity [2]. Since there is no cure for OA, exercise, education, and medication are considered to be cornerstones of its treatment [3,4].

Although patients with knee and hip OA generally tend to avoid physical activity [5], physical exercise is one of the most effective and recommended treatment modalities [3,4]. Exercise therapy, generally provided by a physical therapist, is a regimen of physical activities with the aim to change patients' lifestyle behavior and improve patients' overall function [6]. Therapeutic exercise therapy consists of strengthening, aerobic, flexibility, and/or functional exercises. Multiple studies have demonstrated the beneficial effects of exercise therapy in patients with knee and hip OA. Exercise therapy has positive effects on pain perception and self-reported physical function [7,8]. However, therapeutic exercise therapy is labor-intensive, costly, and often not covered by the health insurance, especially over the long term. So, although helpful, physical therapy is not accessible for many OA patients. According to current estimates, only 7% of all patients with knee and hip OA who are seen in general practice are actually referred to a physical therapist [9].

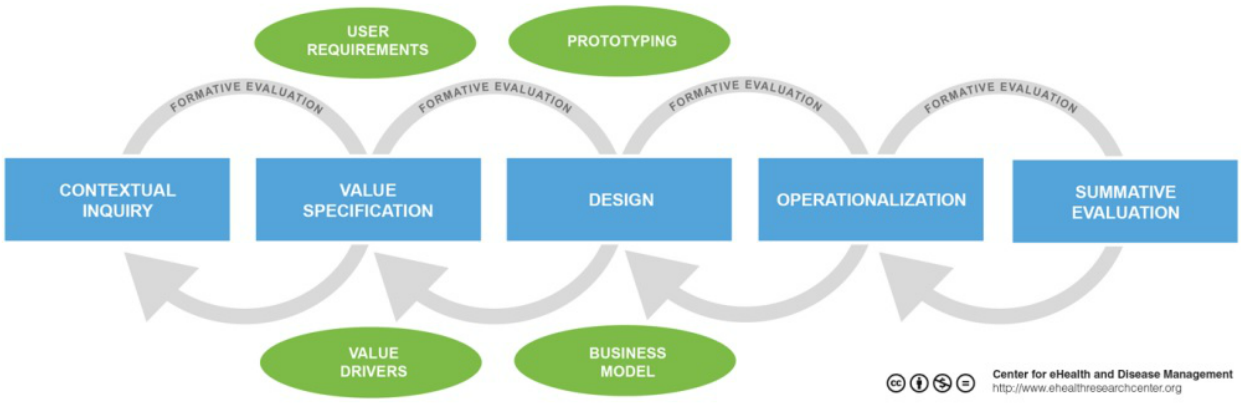
There is a clear need for more feasible and easily accessible strategies in order to regulate therapeutic costs and make exercise therapy attainable for a broader range of OA patients. This can be accomplished through self-management support. Self-management implies individuals' ability to manage the symptoms, treatment, physical and psychosocial consequences, and lifestyle changes inherent in living with a chronic disease [10]. The use of eHealth has the potential to support self-management in patients with OA treatment beyond the walls of the physical therapy practice. Examples include, but are not limited to, Web-based interventions and mobile health interventions that can help to improve patients' health behavior and corresponding health outcomes [11,12]. The 24/7 availability of information may improve treatment compliance, which is critical for the success of physical therapy [13]. Moreover, embedding eHealth within daily practice also has the potential to substitute a part of the face-to-face contacts and alleviate the pressure on health services. Furthermore, eHealth opens up new avenues to reach new patient groups, especially for those who have minimal or no coverage for physical therapy expenses.

Although promising in terms of evidence and accessibility, the adoption of eHealth technologies is disappointing [14]. Embedding eHealth technologies in daily practice is a complex and time-consuming process, more than initially anticipated [15]. So far, eHealth interventions are primarily used outside the health care setting and rarely integrated as part of the treatment. To illustrate, only 1% of all patients in the physical therapy practice use therapeutic-provided eHealth interventions, such as online self-management treatments or online exercises [16]. The uptake and implementation of eHealth innovations in practice is dependent on various factors that can be broadly divided into 4 categories: (1) characteristics of the technology itself, such as ease-of-use and quality of the intervention; (2) characteristics of the end-users, such as perceived usefulness, perceived support from family/colleagues, skills, and knowledge; (3) characteristics of the organization, such as formal endorsement and costs; and (4) policy and legislation, such as privacy issues and reimbursement schemes for eHealth services [17].

The success of eHealth is hampered by insufficient attention paid to abovementioned determinants during the development process. The majority of eHealth technologies is created through ad-hoc procedures without a thoughtful approach [18]. High rates of non-usage and implementation difficulties are a normative phenomenon in eHealth [9,14,19]. The peripheral position of end-users and inadequate input of stakeholders lead to a mismatch between technology and context, which explains why eHealth does not reach its full potential in practice [20]. The involvement of stakeholders—such as health care providers, policy makers, and health insurers—provides direction for the development of eHealth technologies. Co-creation, the engagement of users and other stakeholders throughout the development process, is an important strategy in order to meet the values and needs of stakeholders.

The Centre for eHealth Research and Disease Management (CeHRes) road map is a development approach in which co-creation plays a central role [20]. This CeHRes road map anticipates the needs and values of stakeholders and consists of 5 steps (Figure 1). In this article, we employed the CeHRes road map to develop a new blended intervention for patients with knee and/or hip OA. This intervention, which will be a combination of eHealth and face-to-face care, will be integrated into daily physical therapy practice. This proposed program aims to promote a physically active lifestyle among patients with knee and hip OA. The objective of this study was 2-folded. The first aim of this study is to develop a human-centered eHealth physical activity intervention that matches the values of users and that can be implemented in the daily routine of physical therapists. The second aim was to investigate the feasibility through interviews and a pilot study. To our knowledge this is the first study investigating a blended exercise therapy intervention for physical therapists.

Figure 1. CeHRes road map.



Methods and Results

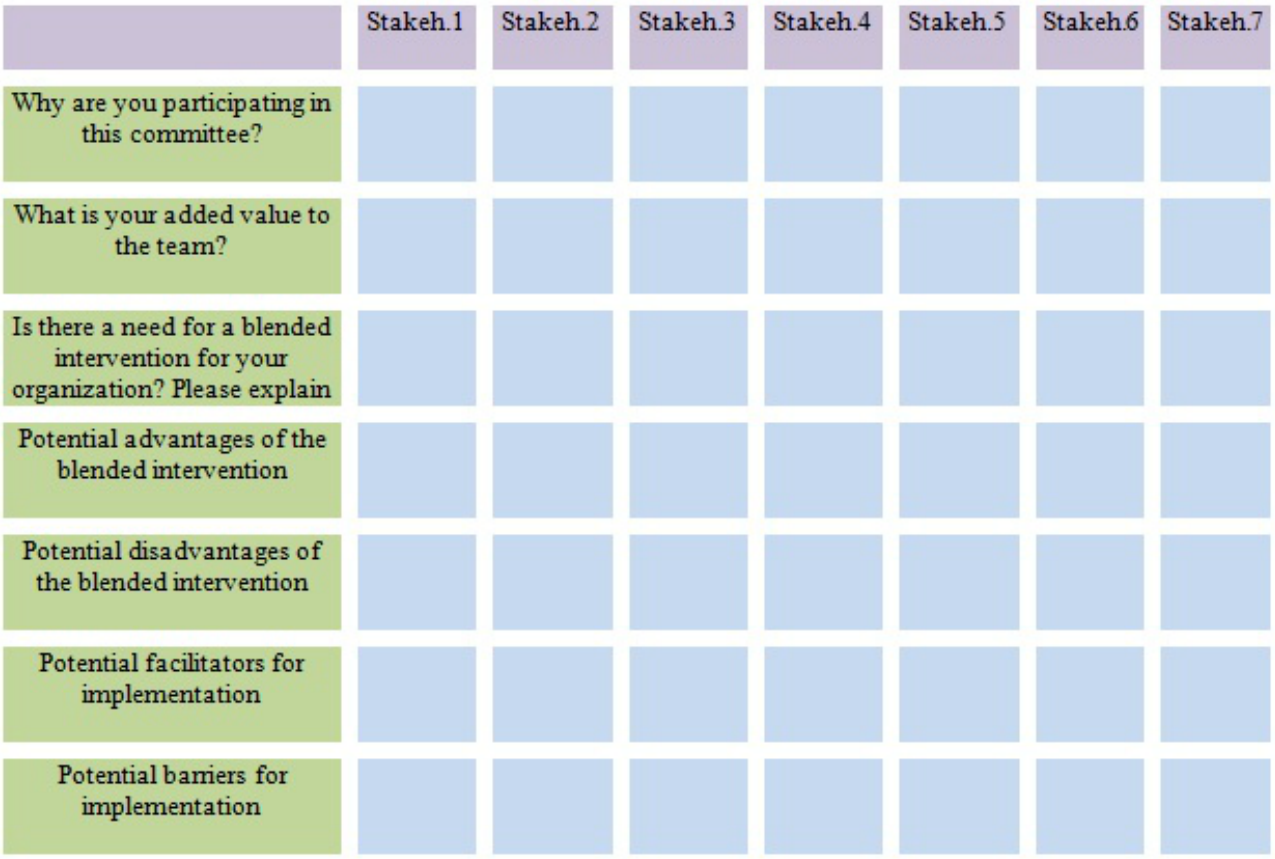
In order to enhance clarity and optimize the execution of each step, we have chosen to present the methods and results sections together. In the following section, we describe the first 3 steps of the CeHRes road map, namely the contextual inquiry, value specification, and design. We present a pilot study on the feasibility of the blended intervention. The first 3 stages of the CeHRes road map and pilot study provide the basis for steps 4 (operationalization) and 5 (summative evaluation), which will be conducted in a later phase of the project. The study has been approved by the Medical Ethical Committee of the St. Elisabeth hospital Tilburg, the Netherlands (Dutch Trial Register NTR4224).

Contextual Inquiry and Value Specification

Methods

During the contextual inquiry and value specification we aimed to establish stakeholders' most important needs, values, and requirements with respect to our to-be-developed blended intervention. The input of this phase was mainly based on another project, executed by the same authors [21]. In this previous work, we developed and evaluated the Web-based intervention Join2move. Join2move is a self-guided intervention and contains automatic functions without human support. The 9-week program is directed at increasing the level of physical activities in a time-contingent manner (fixed time points). More information about the intervention and used methods can be found elsewhere [21]. For the development of our blended intervention, e-Exercise, a focus group among 7 physical therapists was conducted. Physical therapists, who had an extensive experience in the field of OA, were recruited through the website of The Royal Dutch Society for Physical Therapy to participate. The focus group was facilitated by 2 moderators (CK and DB) and lasted approximately 120 minutes. During the focus group, we used a topic guide that contained questions related to the content needs of the intervention, reimbursement, and frequency of face-to-face contact. The focus group discussion was audio-recorded and subsequently summarized. Summarized texts were subsequently read and discussed between 2 reviewers (DB and CK) to gain an overall understanding of the needs and perspectives with respect to the e-Exercise intervention. Furthermore, an implementation committee was formed with different stakeholders. The stakeholder committee consisted of patients with knee and/or hip OA, the Royal Dutch Society for Physical Therapy, 2 rehabilitation centers, the Dutch arthritis foundation, an eHealth entrepreneur, and a health insurer. The committee meetings were held 3 times and were led by the last author (CV). At each meeting, stakeholder members were encouraged to discuss and share their thoughts about the development and implementation process of the blended intervention. The results from these discussions provided direction for further development of the blended intervention. We created a matrix in order to summarize and analyze needs and perspectives of the individual committee members (Figure 2).

Figure 2. Stakeholders' needs and perspectives.



Results

Physical therapists in the focus group indicated that a blended intervention will be a useful instrument in the treatment of OA patients. The 24/7 availability of information and exercises, the possibility to extend the physical therapy treatment in the home environment of the patient, and the potential to enhance the adherence of home exercises were mentioned as possible advantages. On the other hand, the fact that the proposed blended intervention aims to substitute conventional visits may lead to reduced revenues per patient. According to physical therapists, this lack of financial incentive was seen as a potential barrier to use the proposed intervention in practice. The results in the matrix, which represent the stakeholders' needs and perspectives with respect to Join2move, showed positive attitudes toward the to-be-developed blended intervention. As a stakeholder from a rehabilitation institute stated: "Patients will benefit from the blended intervention because it is cheap, independent of time or place, and promotes self-management in the home environment of OA patients." Another facilitator for implementation is the potential to reduce treatment costs. An employee of a health insurance company summarized this by saying: "The proposed blended intervention will possibly result in lower costs since the average number of sessions will be decreased. This will lead to a cost reduction of the OA treatment." The patients were also positive. They had a positive attitude toward the idea that eHealth will be an integrated part of their treatment, especially for information and education purposes.

Design

Methods

E-Exercise is a combination of (1) visits with a physical therapist, and (2) a Web-based physical activity intervention. The technical functionality of the Web-based part is based on a previously developed physical activity intervention [22]. This initial Web-based intervention contained only self-directed features without the integration of physical therapy sessions. To investigate whether and how the initial Web-based intervention fits the day-to-day requirements and routines of physical therapists, different content scenarios were presented during a second focus group session with physical therapists. These scenarios concentrated on several themes, such as the number of face-to-face visits, extent of (online) interaction between patient and physical therapist, and website content such as videos and design and education topics. Results were used to change the first Web-based intervention and create the blended intervention e-Exercise. This first blended version of e-Exercise was then tested in a pilot study.

Results

Over the course of a half year, a team of experts from NIVEL developed the e-Exercise program. The starting point of the development process was a previously developed Web-based exercise intervention [23] and the Dutch guidelines for physical therapists [24]. The intervention is delivered over a period of 12 weeks. During the 12 weeks, patients receive 4 face-to-face sessions with a physical therapist and are supposed to complete 12 online assignments (Figure 3). The physical therapists were

encouraged to follow a fixed treatment protocol. The website has a portal for both patients and physical therapists and contains text- and video-based information. The core element of the website activities is the promotion of moderate physical activities, such as cycling, walking, or swimming in the home environment of patients. Every week, automatic generated physical activity exercises are posted on a password-secured website in which a self-chosen physical activity is gradually increased in a time contingent manner (ie, fixed time points). Time-contingency means that physical activities are increased on fixed time quotas rather than guided by OA-related symptoms such as pain and fatigue. This strategy is derived from the behavioral graded activity intervention and concepts of operant conditioning [25]. The e-Exercise home page is shown in Figure 4. Illustrative screenshots of the e-Exercise website are presented in Multimedia Appendix 1.

Figure 3. Overview of the 12-week e-Exercise treatment.

Figure 4. The e-Exercise home page.

Pilot Study

Methods

Study Design and Objective

This pilot study employed a multicenter 1-group design. The purpose was to evaluate the feasibility of the e-Exercise treatment in the daily practice of physical therapists.

Procedures and Participants

Physical therapists working in a private practice were recruited through the website of the Royal Dutch Society for Physical Therapy and the social network of the authors. Eventually, 8 physical therapists were included in the pilot study. All participating physical therapists received a half day of training about the study procedures and how to use e-Exercise in their practice. Eligible patients, who visited a participating center during the study period, were enrolled by the physical therapists. Enrollment started on March 3, 2014, and ended May 6, 2014. Participants were suitable for inclusion if they (1) were aged 40 to 80 years and (2) had the diagnosis OA of the knee and/or hip according to the clinical criteria of the American College of Rheumatology [26]. Participants were not suitable if they (1) were on a waiting list for a hip or knee replacement surgery, (2) had contraindications for physical activity without supervision, (3) had a physically active lifestyle, (4) participated in a physical therapy for OA and/or physical activity program in the last 6 months, (5) had no access to the Internet, and (6) were unable to understand the Dutch language. Interested patients who were willing to participate and met the eligibility criteria were sent an information letter about the study and an informed consent form. Once written informed consent was obtained, participants were invited to fill out an online baseline questionnaire. After baseline completion, participants were included in the study.

Feasibility

Feasibility measures included website usage, user satisfaction with the website, and recruitment rates of participants within each practice. Program use was measured by the number of modules completed. Based on a previous study [21], we considered the completion of 7 out of 12 modules as feasible. User satisfaction was measured through the System Usability Scale (SUS) [27]. For this development study, an SUS score of 51 points or more was considered feasible [28]. Moreover, participating physical therapists and participants were invited for interviews to learn about their experiences with e-Exercise. Semistructured interviews were conducted on a subsample of 5 physical therapists and 4 patients. Interviews were audio-recorded and transcribed with interviewee's permission. An interview guide with open questions was employed to provide structure to the interviews (see Multimedia Appendix 2). Transcribed texts were read and thematic trend analysis was conducted to identify, analyze, and report recurrent patterns. Themes were discussed by CK and DB to gain an overall understanding of the usability and user satisfaction.

Results

Feasibility

A total of 8 eligible OA patients were included in the pilot study by the 9 participating physical therapists. Patients were on average 62 years old, had 1 or more comorbidities (88%), and most of them were female (75%). None of the participants withdrew from the study. An overview of the sample characteristics is presented in Table 1. Overall, patients and, to a lesser extent, physical therapists were satisfied with the e-Exercise intervention. Results from the system usability scale among patients revealed an average score of 79 points (SD 8.7) on a 100-point scale questionnaire, which can be considered as a good score [28]. Usability scores from physical therapists were considerably lower, namely 64 (SD 7.7). This rating can be interpreted as "fairly" good [28]. Login-analyses showed that 6 out of the 8 patients completed more than 7 of the 12 modules. Over the 12-week intervention period, patients visited the website 33 times on average. Prior the study, we intended to recruit 2 patients per participating physical therapist. However, during the 10-week enrollment period, only 5 of the 8 physical therapists recruited 8 patients in total. Physical therapists reported that e-Exercise is only suitable for a small subset of patients. Most of the patients with knee and hip OA prefer traditional face-to-face treatments over the blended intervention or did not meet the study inclusion criteria. One physical therapist said: "Most of the patients with knee or hip osteoarthritis that I have seen were not interested in participating in e-Exercise because they preferred face-to-face guidance. Other patients did not have a computer or had an already physically active lifestyle."

Overall, interviewees were satisfied with the intervention. One patient summarized this sentiment by saying: "I have told many friends and family that this is a great program because the program motivates you to perform exercises in your own time. I would therefore definitely recommend e-Exercise to others." Physical therapists also expressed positive feedback regarding the content of e-Exercise. To cite 1 therapist: "I am especially pleased with the information about osteoarthritis provided by the videos. More insight into the disease and the role of pain is important prerequisite to encourage a physically active lifestyle." Although physical therapists were generally satisfied, they stressed that e-Exercise must be adapted for suitable integration into practice. As 1 physical therapist commented: "The program provides no insight [into] which modules patients receive. This was truly a downside of the program because I had little or no control over patients' progress." It was also reported by some patients that they liked the effective approach of e-Exercise. One patient commented: "I liked the effective approach of the intervention. You need only a few face-to-face treatments to get on track. The provision of weekly physical therapy sessions is not useful because you have to exercise yourself."

Table 1. Baseline demographics and characteristics OA patients.

Participants (N=8)		
Gender, n (%)		
	Female	6 (75)
	Male	2 (25)
Mean age, y (SD)		61.88 (14.53)
Location OA, n		
	Knee	4
	Hip	3
	Both	1
Duration of symptoms, n		
	< 1 year	2
	1-3 years	1
	3-7 years	2
	≥7 years	3
Education, n		
	Low	2
	Middle	2
	High	4
Comorbidity, n		
	None	1
	1	4
	≥2	3

Discussion

Principal Findings

Provision of blended care requires a harmonious integration of technology into practice, combining complementary face-to-face treatments with eHealth technology. Implementing a blended intervention into health care is a complex process that changes existing routines, relationships, and budgets. Developers and researchers have to anticipate these implementation difficulties. While research supports the effectiveness of health technology, health care professionals often lack the time, skills, and resources to integrate eHealth into their daily practice. Input of end-users and other stakeholders throughout the development process is a prerequisite for the successful implementation of blended interventions into practice [20]. The aim of this study was to develop and investigate the feasibility of a blended exercise therapy intervention for patients with knee and hip OA that can be implemented in the daily routine of physical therapists.

The involvement of patients, physical therapists, and other stakeholders was extremely valuable throughout the development process. The first 3 phases of the CeHRes road map yielded unique insights into different needs and values of end-users and various stakeholders. Steps from the CeHRes model were not purely sequentially executed but involved a continuous process. For instance, the identification of needs and problems was mainly derived from experiences with a previous eHealth project, rather than a separate phase in the current project. The results from the post-pilot interviews demonstrated that e-Exercise is feasible in the treatment of patients with knee and hip OA. In line with the findings from the study by Pietrzak et al [29], participants were positive toward the use of eHealth in the treatment of OA. Users considered the usability of e-Exercise as “good.” Interviews with physical therapists and patients revealed a beneficial impact on the organization process of care. The possibility to stimulate exercises in the home environment and to enhance exercise adherence were cited as major advantages. However, the inability to monitor patients’ progress between consultations seemed to be a drawback. Monitoring was therefore added in the latest version of e-Exercise. Another sign that demonstrated the feasibility of e-Exercise were the usage rates. Of the 8 participants, 6 completed more than 7 of the 12 modules. These usage rates can be considered reasonably high when compared with a previous study [21].

The visit-based method of recruitment was challenging. Over the 10-week enrollment period, we intended to recruit 2 patients per participating physical therapist. However, only 8 eligible patients were recruited by 8 physical therapists. Others have reported similar challenges with the recruitment of patients [30-32]. The lack of remuneration may have contributed to the disappointing recruitment rates since there is no financial incentive to adopt e-Exercise in practice. On the contrary, the

use of e-Exercise might even lead to reduced revenues per patient. Another possible explanation for the poor recruitment rates is the small pool of eligible OA patients. Primary care data from the Netherlands shows that only 2% of all patients seen in the physical therapy practice have OA [33]. General practitioners, the gatekeepers of the Dutch health care system, have a strong influence on the influx of patients into physical therapy setting. General practitioners should therefore be informed of the availability and possibilities of blended interventions. This might influence the referral behavior of general practitioners positively.

Limitations

The findings of the pilot study need to be interpreted in light of several limitations. The small number of participants and the absence of a control group are major limitations of the current study. Moreover, the generalizability might be limited by the self-selected sample in this study. Obviously, included physical therapists are techno-enthusiasts who are more willing to adopt technology in their practices than are others.

Conclusions

Results from this study are valuable to set up a follow-up study to compare e-Exercise with usual physical therapy. We plan to conduct a larger, adequately powered, randomized controlled trial to investigate the effectiveness (including cost effectiveness) of e-Exercise [34]. The recruitment of patients was a true challenge in this study. We therefore need to pay extra attention to the recruitment process and find additional avenues to increase recruitment rates for the randomized controlled trial. Given the 1:1 recruitment ratio of this pilot study, we aim to recruit at least 200 physical therapists. We also plan to use strategies to encourage physical therapists to include more participants, to engage general practitioners in the recruitment of patients, and to extend the inclusion period of patients.

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Conflicts of Interest

None declared.

Multimedia Appendix 1

Screenshots website.

[PDF File (Adobe PDF File), 572KB - resprot_v5i1e32_app1.pdf]

Multimedia Appendix 2

Interview guide.

[PDF File (Adobe PDF File), 228KB - resprot_v5i1e32_app2.pdf]

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Abbreviations

CeHRes: Centre for eHealth Research and Disease management
OA: osteoarthritis
SUS: System Usability Scale

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Protocol

Understanding User Reactions and Interactions With an Internet-Based Intervention for Tinnitus Self-Management: Mixed-Methods Process Evaluation Protocol

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Abstract

Background: Tinnitus is a common medical symptom that can affect an individual's emotional and functional quality of life. Psychological therapies are acknowledged as beneficial to people with tinnitus; however, such therapies are not always readily accessible. With their global reach, automated Internet-based interventions have the potential to reduce the disparity in access to psychological support that people with tinnitus currently experience. However, the evidence on the acceptability and efficacy of these interventions is lacking. Process evaluations that develop an in-depth understanding of how users experience these interventions provide an essential first step when evaluating complex psychological interventions.

Objective: To describe the protocol for a study that will explore past, current, and new users' reactions to and interactions with the Tinnitus E-Programme, an Internet-based intervention for the self-management of tinnitus.

Methods: Two parallel mixed-methods studies will be carried out with 2 different populations. Study 1 will use an online survey to gather past and current users' views of the program. Study 2 will recruit new program users to take part in an interview and complete a relaxation log to explore how well they were able to implement the skills they learned during the program in their everyday lives. The findings from both studies will be triangulated to develop an in-depth understanding of the program's mechanisms of impact and identify any implementation or contextual factors that strengthen or impede its delivery and functioning.

Results: Study 1 is open for recruitment with a projected completion in June 2016 and Study 2 was completed November 2015. At the time of submission, 36 participants have been recruited to Study 1 and 12 participants have taken part in Study 2.

Conclusions: Findings will inform the optimization of the Tinnitus E-Programme and guide future evaluation work to assess the program's effectiveness as a therapy for people with tinnitus.

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KEYWORDS

Tinnitus; health; internet; program acceptability

Introduction

Background

Tinnitus (ringing in the ears) affects approximately 10%-18% of the population [1-3] and is characterized by a conscious perception of a sound without a corresponding external source. Tinnitus can significantly affect an individual's quality of life, leading to emotional strain (eg, irritation, depression, frustration, anxiety), sleep disturbances, concentration difficulties, and disruptions to social and work life [4-6].

In the absence of a definitive biomedical cure, current health care strategies focus on supporting people to manage their tinnitus to ultimately reduce the tinnitus percept and associated psychological distress [7]. Currently, access to psychological therapies, such as cognitive behavior therapy, for people with tinnitus is limited [8-10], with such specialist psychological services generally being reserved for those with the greatest need [7]. Internet-based interventions, with their global reach, have the potential to reduce this disparity and improve access to psychological support for people with tinnitus. They also provide an alternative for those unable or unwilling to access traditional face-to-face psychological services [11,12].

There is evidence to suggest that Internet-based interventions are effective for reducing tinnitus distress and psychological comorbidity while improving quality of life [13-16]. However, the current evidence-base has focused on therapist-guided interventions, which lack the scalability necessary for equitable access. On the other hand, the evidence-base for unguided (or automated) Internet-based interventions is limited and less clear [14,17,18]. One such example is the Tinnitus E-Programme, an Internet-based intervention to support tinnitus self-management that was developed in the United Kingdom [19]. The program comprises several self-management components including: education about tinnitus and its management; information about available resources; training in psychological strategies (ie, relaxation, cognitive restructuring); peer support via an online discussion forum; and self-monitoring of tinnitus outcomes. Its multicomponent nature defines the program as a complex intervention [20]. Although freely available online, we currently know little about how the Tinnitus E-Programme is used, how it works, the circumstances in which it works best, and whom it works best for.

To evaluate the Tinnitus E-Programme, we are guided by the Medical Research Council's guidance on developing and evaluating complex interventions [20] that emphasizes the importance of carrying out adequate pilot and feasibility work prior to a definitive randomized controlled trial. Interventions should be tested using a phased approach whereby a series of pilot and exploratory studies address any key uncertainties in the intervention design. Development and evaluation stages are iterative, with researchers moving back and forth between each stage. Any intervention modifications and future evaluation work is thus informed by an evolving evidence-base produced by these pilot studies. Without adequate development and piloting work, interventions are likely to be weaker and difficult to evaluate [20].

A useful first step when evaluating developed interventions is to carry out a process evaluation. This can provide information on the (1) implementation, (2) mechanisms of impact, and (3) contextual factors that influence the delivery and outcome of the intervention [21]:

1. Implementation is concerned with what is delivered in practice and the structures and resources required for successful implementation [21] and is typically conceptualized in terms of fidelity (ie, was the intervention developed and used as intended?), dose (ie, how much of the intervention was delivered and received?), reach (ie, to what extent did the intervention reach its target audience?), and enactment (ie, to what extent was the knowledge or skills participants acquired during the intervention applied to everyday life?) [22-24]. In the context of Internet interventions, usability testing is essential for ensuring that the intervention performs as intended and identifying and eliminating any barriers to easy and effective use by its target population [25]. Exploring intervention usage or attrition can also provide useful implementation insights [26].
2. Mechanisms of impact is concerned with how the intervention components—and a user's interactions with them—lead to the desired changes in outcome [21]. That is, what are the mechanisms through which Internet interventions work (ie, how they work) and the factors that are essential for their success (ie, what makes them work)? Qualitative methods can be particularly helpful for exploring relatively unknown mechanisms of impact and allow unintended and/or unanticipated intervention consequences to be explored [27]. This may include identifying negative intervention outcomes or benefits not initially anticipated by the intervention developers or evaluators. For example, in a mixed-methods evaluation of psychological therapies for multiple sclerosis, Dennison et al's [28] qualitative interview findings uncovered a disparity between participants' perceptions of what the therapy changes were and the predetermined outcomes measured in the parallel efficacy trial.
3. Context is concerned with how external factors may strengthen or impede the delivery and functioning of the intervention [21]. Such external factors may include preexisting circumstances, skills, resources, and attitudes of the target population. A thorough understanding of the intervention context is helpful for explaining any variability in intervention outcomes [28,29].

This study will carry out a process evaluation of the Tinnitus E-Programme to further our understanding of the program's mechanisms of impact and identify any implementation or contextual factors that strengthen or impede its delivery and functioning. Most process evaluations have been carried out on people who were recruited offline and are using the intervention for the first time as part of a research study. This reduces the findings' external validity and relevance to real-world practice [30-32]. This study will recruit users of the live program, as well as people with tinnitus who have not used the program previously. Mixed methods will be used to develop an in-depth understanding of the perspective of the target user [33]. The findings will inform the optimization and future evaluations of the program, as well as the development of other similar internet-based self-management interventions.

Aims

To explore past, current, and new users' reactions to and interactions with the Tinnitus E-Programme. The specific aims are to explore:

1. The acceptability and usability of the program (implementation, context);

2. How users engage with the program (implementation, mechanisms of impact, context);
3. Users' perceptions of the processes and outcomes of the program (mechanisms of impact, context);
4. User enactment of the relaxation skills learned in the program (implementation, mechanisms of impact, context).

Methods

The Intervention

The Tinnitus E-Programme [34] is a 10-week Internet-based self-management intervention for tinnitus. It was developed by a hearing therapist/psychotherapist in private practice and was launched in 2009. It is live online and free to access without registration. The website currently receives approximately 1000 visits per month. The program includes: (1) downloadable information resources to provide education about tinnitus and its management; (2) training/rehearsal for psychological strategies, including relaxation and brief cognitive restructuring skills training; (3) online discussion forum to provide social support from peers and lay and professional moderators; (4) self-monitoring of tinnitus distress using the Tinnitus Handicap Inventory [35]; and (5) information about available resources, including book references and hyperlinks to other websites or services. Educational topics covered by the information resources include the mechanisms of tinnitus, stress and its management, attention focus, and negative thinking. Several behavior change techniques are also used to promote relaxation behavior (eg, goal setting, action planning, behavioral practice/rehearsal). Further information about the Tinnitus E-Programme's specific components, techniques, and mode of delivery can be found elsewhere [19].

Program content is delivered across 6 weekly modules, followed by a 4-week maintenance period where users are asked to continue the daily relaxation goals set in the previous period. No additional intervention content or support is delivered during this maintenance period. A recommended program structure is given; however, users have free choice regarding which components they access and in what order they access them. The express aim of the program is to reduce tinnitus distress, but the precise mechanisms by which this change should occur are not yet established.

Paradigm and Design

This research will adopt pragmatism [36] as its overarching methodological paradigm. Pragmatism is primarily concerned with the consequences of research. Unlike other paradigms, such as postpositivism and constructivism, pragmatism is not tied to one particular epistemology or data collection method (ie, qualitative or quantitative). Rather, methods are chosen based on "what works," that is, their ability to successfully answer a particular research question.

Consistent with this approach, 2 parallel mixed-methods studies will be carried out with 2 different populations to evaluate the program from multiple perspectives. This design will allow triangulation of research data and methods that will generate and compare complementary perspectives and contexts. The intention is that the use of both qualitative and quantitative research methods and more than 1 study population will provide a more complete, in-depth, and valid understanding of the phenomenon than if only 1 method or population was used [36,37]. Mixed methods have been used successfully for process evaluations [21] and evaluations of digital interventions [26,38].

Study 1 will explore how past and current users react to and interact with the program in the real-world, outside of a research context. Due to technical limitations of the program, it is not possible to monitor actual program usage. Therefore, an online survey will be used to gain self-reports of how users interacted with the program, as well as users' reactions to the program. A convergent mixed-methods design [36] will be used in which qualitative and quantitative methods are implemented simultaneously and given equal weight, but the data will be analyzed separately. The online survey will use open (ie, qualitative) and closed (ie, quantitative) questions to elicit users' views. Specifically, a data-validation variant of this mixed-methods design will be used [36] in which the qualitative data is used to validate and elaborate on the quantitative data.

Study 2 will recruit a cohort of individuals with tinnitus who have not previously used the Tinnitus E-Programme in an attempt to gather more in-depth, timely, and diverse views and experiences. Participants will complete the program for the first time and take part in a semistructured interview. Participants will complete a relaxation log to explore the extent to which they enacted the relaxation skills learned in the program and any barriers to doing so. An adapted version of an embedded mixed-methods design will be used [36] in which both the qualitative and quantitative relaxation log data collection and analysis is embedded within an overall qualitative research design. As such, the relaxation log data will be secondary to the qualitative data and will be used to enhance understanding of the qualitative interview findings.

The findings of the 2 mixed-methods studies will be triangulated in an overall interpretation. This research has ethical approval from the University of Nottingham Research Ethics Committee (Reference Number: Q11122014 SoM NIHR RHA QEST).

Study 1: Online Survey With Current and Past Users

Participants

Participants in Study 1 will self-select, based on their own judgements of whether they meet the following inclusion criteria: (1) adults aged 18 years and over, (2) ability to read English, (3) access and ability to use the Internet, and (4) have visited the Tinnitus E-Programme website or used the program. Participants may have accessed the program anytime over the last 6 years since the program was launched. There will be no exclusions regarding length of time since starting the program in order to maximize recruitment for this very specific population. The program does not specify any inclusion criteria, as the intention is that it is suitable for everyone with tinnitus. In keeping with this, there are no exclusions regarding tinnitus duration, severity, or co-morbidities in an attempt to recruit all potential users.

Recruitment

Past and current program users will be invited to take part in an online survey hosted on SurveyMonkey. Advertisements will be posted on the Tinnitus E-Programme website and online discussion forum, along with the participant information sheet. The survey will also be advertised via social media and national charities in an attempt to reach those who no longer interact with the program or website. Email invitations and the participant information sheet will also be sent to those who registered with the program website or online discussion forum. Sample sizes for similar descriptive online survey studies have been between 50-249 individuals [39-42]; therefore, a sample size of above 50 will be deemed acceptable. The survey will be closed after 3 months or until at least 50 participants have been recruited.

Online Survey: Development and Piloting

The initial survey design was informed by the study rationale, relevant literature, and the comprehensive intervention description developed previously [19]. The survey focused on the information resources (ie, education about condition and management, information about available resources, training/rehearsal for psychological strategies), relaxation exercises (ie, training/rehearsal for psychological strategies), Tinnitus Handicap Inventory (ie, self-monitoring of condition), and online discussion forum (ie, social support).

The survey uses a mix of closed and open questions concerned with: (1) reasons for participating or not participating in the program, (2) how the program was used, (3) usability of the program, (4) acceptability of the individual program components, and (5) benefits derived from the program and its impact on tinnitus management. Demographic data will also be collected on gender, age, country of residence, whether English is their first language, presence of tinnitus, tinnitus duration, and tinnitus management strategies used previously or currently.

To assess the acceptability and face validity of the survey, an initial set of survey questions were reviewed by a public and patient involvement (PPI) panel assembled for the purposes of this study. The panel included 4 people with tinnitus and/or hearing loss who were recruited from an established National Institute for Health Research (NIHR) Nottingham Hearing Biomedical Research Unit (NHBRU) PPI panel and 1 voluntary sector representative from the British Tinnitus Association who had experience in writing communication materials for people with tinnitus. Panel members were chosen from a wider established PPI panel, based on their availability and previous experience of reviewing research materials. A focus group was carried out with the PPI panel to gather initial feedback on a paper version of the draft survey. The focus group was attended by the first author and co-facilitated by the PPI manager at NHBRU and an external facilitator who was not involved in the study but was familiar with issues relevant to hearing research. Panel feedback focused on the relevance and ordering of the questions, language used, and appropriateness of question type (ie, closed or open). Following the focus group, the survey was uploaded onto SurveyMonkey, and this online version was circulated to the PPI panel via email for additional comments. The panel was satisfied with the online version and no further amendments were made.

The final online survey was subsequently piloted with 3 Tinnitus E-Programme users recruited from the program's online discussion forum. These participants completed the online survey and answered 4 additional questions about the survey length and relevance of the questions and closed-question answers. Informed consent was gained from these pilot participants who were told that their answers may or may not be used in the final analysis, depending on the outcome of the pilot.

All 3 participants reported that the survey took less than 30 minutes and "just the right amount of time" to complete. One participant suggested that it would be helpful to add a free-text comments box next to some of the closed questions to allow people to clarify their answers. The same participant also suggested adding a "cannot remember" option for the questions regarding program usage (eg, did you use the online discussion forum?) for those who used the program a long time ago. Both of these changes were made to the final survey. As these amendments were minor, the pilot data was retained for inclusion with the main study. A copy of the final survey can be found in Multimedia Appendix 1.

The online survey is anonymous to encourage participation and only 1 submission per computer will be allowed. Participants will be given a 14-day period in which they can request to have their answers deleted. After this period, their answers will be downloaded onto university servers and cannot be deleted. Participants will be asked to provide a security word as part of the survey and will be asked to recite this for data identification purposes should they wish to withdraw their data from the study.

Analysis

Answers to closed questions will be analyzed in IBM's SPSS Statistics 22 using descriptive statistics, including frequencies and percentages, and each statistic carried out on complete data only. Answers to the open questions will be analyzed separately using inductive thematic analysis [43] and analysis informed by guidelines for establishing validity in qualitative research [44,45]. QSR's NVivo 10 qualitative data analysis software will be used to provide an audit trail.

First, the 3 coders (KG, MS, DH) will familiarize themselves with the data through repeated reading of the survey answers. Second, KG will utilize line-by-line coding, a technique from grounded theory [46], in which each line of your transcript is coded. This ensures the coder remains open to the data and that subtle nuances in it are not missed. Codes will be kept close to the text and participants' own language will be used wherever possible. KG will develop a coding manual that will list all codes, including descriptions and example quotes from the text [47]. The coding manual will improve the rigor of the research while also providing an audit trail for analysis decisions.

Third, at least 1 other coder (MS, DH) will independently apply the coding manual to all transcripts to clarify ambiguous codes, remove duplicate codes, and identify data that did not fit the coding scheme. Coding will be compared and discussed between coders and subsequent modifications made to the coding manual.

Fourth, coders will collectively organize these codes into overarching themes and the coding manual will be updated accordingly. The constant comparison method [48], a grounded theory technique, will be used to compare codes across different participants, contexts, and situations. Disconfirming case analysis [45] will be used to actively identify data that does not fit with the identified themes. The final interpretations will be reviewed and agreed by all authors. Participant quotes will be used in the final write-up to illustrate the themes.

Consistent with a data-validation variant of the convergent mixed-methods design [36], the qualitative findings will be used to validate and elaborate on the quantitative data.

Study 2: Interviews and Relaxation Log With New Users

Participants

Participants in Study 2 will self-select, based on their judgement of whether they meet the following inclusion criteria: (1) adults aged 18 years and over, (2) ability to read English, (3) access and ability to use the Internet, (4) have self-reported tinnitus, (5) reside in the United Kingdom, and (6) have not previously used the Tinnitus E-Programme. Again, as the program is meant to be suitable for all tinnitus users, participants were not excluded based on any tinnitus-related characteristics.

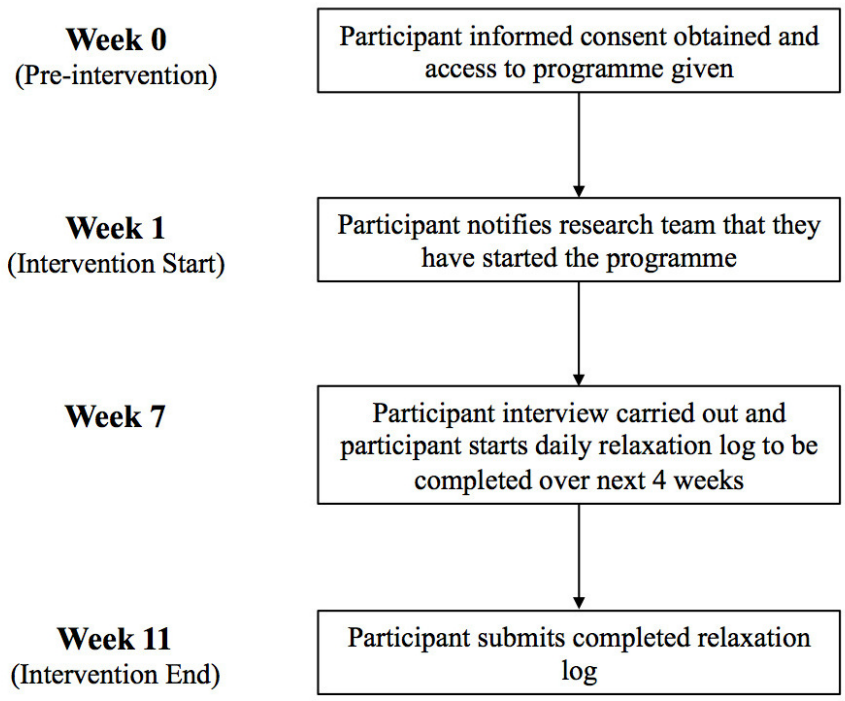
Recruitment and Procedure

A purposive sample of people will be chosen from the NHBRU research database, which includes approximately 900 UK residents with tinnitus who have agreed to be contacted about research. Maximum variation sampling [49] will be used to ensure that a diverse sample with different demographics (eg, gender, age) is chosen. As recruitment progresses, targeting will become more specific as participants with certain characteristics (eg, hearing loss, short tinnitus duration, younger age) are actively sought out to fill any demographic gaps in the current sample. An email invitation, together with a participant information sheet, will be sent to selected database members by a member of the research team.

The procedure for Study 2 is illustrated in Figure 1. Once participants have expressed an interest in the research, the researcher will gain their informed consent using a paper or electronic consent form. Recruited participants will then be sent the hyperlink to the Tinnitus E-Programme and asked to notify the researcher once they start using the program. An interview will be organized for approximately 6 weeks after their start date. During this time, participants should have sufficient time to complete the first 6 sections of the program and be progressing into the maintenance phase. One week before their interview date, participants will be emailed a set of sample interview questions, a hyperlink to their online relaxation log, and instructions on how to complete the log. Participants were sent a set of sample interview questions to encourage transparency with the interview process and improve recall by giving participants time to think about the different topic areas and revisit the website if needed [50].

Interviews will be held no later than 8 weeks after the participant's start date to explore how acceptable the 6-week timeline is and to ensure maximum recall of intervention experiences. Participants will be asked to complete a daily relaxation log on paper or online over the following 4 weeks. Email reminders will be sent to those who have not yet started the program, organized an interview date, or completed their relaxation log. Recruitment will cease once data saturation has been reached for the interviews; that is, when no new themes are emerging [51].

Figure 1. A flow chart showing the procedure for Study 2.



Interviews

An interview guide was developed that was informed by the literature, intervention coding, and study rationale. Specifically, the interview questions are concerned with how people used the program; reasons for any nonusage; experiences of using the program; expectations of the program; usability and acceptability of the program, as well as its individual components; benefits derived from the program; and suggested improvements to the program. The interview guide can be found in Multimedia Appendix 2. The interview guide was reviewed by the NHBRU PPI panel and piloted with a previous user of the Tinnitus E-Programme. No modifications resulted from this process.

Demographic data—including gender, age, ethnicity, and tinnitus duration—will also be obtained. Interviews will be carried out by the first author, a health psychologist and PhD student experienced in qualitative interviewing who was not involved in the Tinnitus E-Programme's development. Interviews will last no longer than 1 hour. Participants will be given the choice of being interviewed in person at the research unit, over the phone, or via video chat. In an attempt to be inclusive, those with severe or profound hearing loss will also be offered the option to be interviewed using text communication methods (eg, instant messaging or email). The audio from the interviews will be recorded using a digital voice recorder and transcribed verbatim. The text from the textual communication methods will be saved electronically.

Relaxation Log

The relaxation log will assess users' enactment of the relaxation goals set by the program during the 4-week maintenance period. An online relaxation log will be created for each participant using Google Sheets, an Internet-based spreadsheet program. Google Sheets will be hosted on the NIHR Google Hub, a secure online file storage system. The relaxation log is in tabular format with 4 columns and 28 rows representing each day of the 4-week period. Users will be required to answer the following 3 questions each day:

1. Did you practice the mind calming breathing exercise 3 or more times today?
2. Did you practice any of the 30-minute relaxation exercises today?
3. Did you use any other parts of the Tinnitus E-Programme today? If yes, please write which parts. If no, please write "no."

There is also a free-text comments box to write any other comments for each day. At the end of week 10, participants will be asked to answer 1 final open question: "Did you practice the recommended relaxation exercises every day? If not, could you tell us about some of the things that made it difficult to do so?"

Participants will have the option of either accessing and completing their online log each day or printing and completing a paper copy. The completed paper copy may be posted or transferred onto the online log. Participants will be provided with instructions for completing their online relaxation log. Relaxation logs will be anonymous, identified only by a unique participant identification code. Each participant will be given a unique hyperlink to access their personal log, and only the participant and researcher will have access to this hyperlink.

Analysis

The interview data and open-question responses from the relaxation logs will be analyzed together using the same inductive thematic analysis strategy outlined in Study 1. The line-by-line coding will begin during data collection to help the interviewer to reflect and learn from previous interviews and refocus future interviews [46]. The quantitative relaxation log data will be analyzed using frequencies and percentages, including complete data only. This quantitative data will provide a secondary and supportive role to the qualitative data and will be used to enhance the qualitative accounts.

Overall Interpretation

The findings from the 2 mixed-methods studies will be triangulated (ie, compared and contrasted) at the discussion-writing stage to produce an in-depth understanding of the program's mechanisms of impact and identify any implementation or contextual factors that strengthen or impede its delivery and functioning. Triangulation will allow the findings from each study to be corroborated and validated [36,37].

Results

At the time of manuscript submission, 36 participants have consented to take part in the online survey in Study 1. Thirty of these participants went on to answer questions about the program. For Study 2, 12 participant interviews have been completed and 6 relaxation logs submitted. Data collection for Study 2 was completed November 2015. Study 1 is open for recruitment and data collection will complete in June 2016.

Discussion

This protocol describes 2 mixed-methods studies to evaluate the Tinnitus E-Programme, an Internet-based intervention for tinnitus self-management. A process evaluation will explore past, current, and new users' reactions to and interactions with the program.

Ultimately, the findings of this research will provide the missing evidence-base that is necessary to guide future optimization and evaluation work for the program. First, the identification of any implementation or contextual factors that impede the delivery and function of the program will help us to decide which amendments need to be made to improve the program's content, usability, and enactment for future users. Second, an in-depth understanding of the psychosocial context in which people with tinnitus interact with the program will provide insight into the circumstances in which the program works best and who is likely to benefit most from it. This can help guide decisions regarding appropriate research conditions and inclusion criteria for future evaluation studies. Third, understanding users' perceptions of the outcomes of the program can guide evaluation choices regarding appropriate outcome measures. Finally, understanding the program's mechanisms of impact can give us an understanding of how the program works and what makes

it work. Such an understanding has wider implications for the management of tinnitus and can also inform the development of other Internet-based programs for people with similar conditions.

Limitations

This research has some limitations or challenges that need to be considered. As registration to the program is not mandatory and users can choose to complete it anonymously, we have no way of knowing how many people have previously used, or are currently using, the live program. This makes it difficult to reliably estimate the sample size and accurately assess external validity for Study 1. It is also not possible to track past and current users who did not register, making this target population potentially hard to reach and recruit. This limitation also means that convenience sampling was the only feasible sampling method, which may introduce a self-selection bias. The current program does not monitor actual program usage, which means that it will not be possible to validate whether participants actually used the program and their self-reported usage. However, the focus of this exploratory study is on the participants' accounts of their usage and reasons for usage or any nonusage.

Participants recruited to Study 1 are likely to represent a particularly motivated and satisfied group of users. Those who chose not to use the program, gained no benefit from the program, or no longer use the program are less likely to take part. Study 2 will provide more diversity as it will introduce a group of people to the dataset with different motivations (eg, to support tinnitus research, looking to benefit from a novel intervention). Once recruited, Study 2 participants will be encouraged to continue onto the interview, even if they did not complete or benefit from the program.

In Study 1, there were no exclusions regarding length of time since completing the program to maximize recruitment for this very specific population. Some of the participants may have completed the program as long as 6 years ago, which may introduce a recall bias.

Conclusions

There are also several strengths of this research. First, the proposed evaluation is being carried out by an independent research team who were not involved in the development of the program. This will minimize any biases that might be present during data collection, analysis, and interpretation. Second, 2 different populations—current or past and new program users—will be studied, allowing us to evaluate the program from 2 different but complementary perspectives and contexts. Combined with the use of mixed methods, this design will provide a more complete, in-depth, and valid understanding of users' reactions to and interactions with the program. Finally, this study will explore users' enactment of the relaxation skills learned in the program. This aspect of intervention implementation is more commonly explored in behavior change research where integrating new actions into everyday life is the ultimate outcome of interventions [52]. Enactment has rarely been studied in research on interventions addressing psychosocial outcomes [22], with most research focusing on dropout or nonusage attrition [17,53,54]. This evaluation will also use mixed methods to relate individual assessments of enactment with user's qualitative accounts of their experiences and reactions to this skills training.

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Conflicts of Interest

None declared.

Multimedia Appendix 1

Online survey for Study 1.

[PDF File (Adobe PDF File), 165KB - resprot_v5i1e49_app1.pdf]

Multimedia Appendix 2

Interview guide for Study 2.

[PDF File (Adobe PDF File), 98KB - resprot_v5i1e49_app2.pdf]

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Abbreviations

NHBRU: Nottingham Hearing Biomedical Research Unit
NIHR: National Institute for Health Research
PPI: public and patient involvement

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Protocol

Combining Persuasive Technology With Behavioral Theory to Support Weight Maintenance Through a Mobile Phone App: Protocol for the MotiMate App

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Abstract

Background: The prevalence of health-focused mobile phone apps available for download increases daily, with weight management apps being among the most proliferative. However, most lack theoretic grounding or evidence of efficacy. There is a significant body of literature which provides evidence for behaviors which are associated with successful weight loss maintenance. Behavioral theory also provides further insight regarding successful behavior change and maintenance.

Objective: We aimed to apply this knowledge to the development of the functionality of an app targeting weight loss maintenance.

Methods: We have subsequently undertaken the development of a persuasive and behavior targeting mobile app (MotiMate) to assist in maintenance of weight loss. MotiMate combines persuasive and behavior change theories in a practical targeted tool through its motivational messages, personalized feedback, and intelligent supportive tools to manage weight, food, exercise, mood and stress.

Results: The development and trial of MotiMate received funding support in May 2014. All 88 volunteers started the trial by December 2014 and were in the process of completing their final visits when this paper was submitted (May 2015). Data analysis is currently underway.

Conclusions: The paper has presented a scientifically informed mobile phone app to support weight loss maintenance. Further evaluation of its efficacy is in progress.

Trial Registration: ANZCTR 12614000474651; https://www.anzctr.org.au/Trial/Registration/TrialReview.aspx?id=366120 (Archived by WebCite at http://www.webcitation.org/6eJeQiKxi).

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KEYWORDS

app; weight maintenance; psychology; coping

Introduction

Thirty percent of the global population is overweight or obese, with this predicted to rise to almost 50% by 2030 [1]. Many people have success in changing their dietary and/or physical activity behaviors to lose weight; however, few successfully maintain their lost weight over the longer term. In order to gain the multiple health benefits of weight loss (such as improved heart health), lost weight must be maintained. A review of The National Weight Control Registry in the US found only 20% of people managed to maintain initial losses after 2 years. Those members reporting maintained weight loss engaged in more

physical activity, monitored their weight regularly and maintained a consistent eating pattern throughout the week [2]. If individuals can successfully maintain their weight loss for 2-5 years then the chance of longer-term success increases greatly [3].

To promote the behavior change needed for weight control, an interactive device such as a mobile phone has the capacity to intervene at critical times or provide ecological momentary intervention (EMI). A review of EMIs using portable devices identified 5 papers that have targeted weight loss [4]. All of these systems were designed to offer behavior change techniques (such as goal setting and monitoring) and keep users motivated, with some focus on the provision of feedback and others giving psycho-education. The review authors concluded that EMI through portable devices may be useful to enhance cognitive-behavioral therapy for weight loss programs. There was limited evidence evaluating the effectiveness of EMI delivered in the absence of other more intensive programs. A separate review focusing specifically on mobile phones suggested that existing systems have underutilized the functionality of modern phones, by relying on short message service (SMS) and not providing immediate two-way feedback, for example [5].

Despite a high number of weight control apps publicly available, few are scientifically derived [6]. Furthermore, despite the fact that theoretically based interventions may be more efficacious for behavior change [7], authors reviewing mobile phone interventions conclude, “a paucity of discussion regarding the health behavior theories or models that provide the basis of intervention” [7].

Behavioral theory can offer several insights and more in-depth understanding of how to achieve successful behavior change when in combination with apps. Conservation of resources and self-regulation theory suggest that an individual has limited capacity to navigate stresses successfully through each day and to do that, people must utilize the appropriate resources [8,9]. These extend beyond an individual’s capacity to control their own behavior to other more tangible resources such as social support and even practical things such as money [8]. The more resources one has in their possession or reach, the more likely that they will be able to use them to avoid stress, overcome barriers, self-regulate and achieve desired behaviors. For example, a person trying to limit intake of fatty snacks may run out of psychological resources to resist temptation (self-regulation) but could then use other resources such as friends to keep motivated (social support). There is evidence that people low in psychological resources to deal with hassles may be more vulnerable to illness [10] which suggests that resources are important for well-being. There are clear intersections between the concept of limited resources and the potential benefits of adaptive coping strategies. For example, coping strategies could become a resource for an individual to use to overcome their challenges [11]. Considering that people may try 6-7 strategies when addressing a hassle [12], it is not surprising that the more coping strategies or resources a person possesses, the more likely it is that they will find one that successfully helps them to sustain their desired behavior change.

The ability to recognize, address and cope with the multiple challenges that come with behavior change is an essential part of long-term success. For weight maintenance, the possible coping scenarios for individuals in different situations are almost infinite. Removing focus from behavioral minutiae toward one’s global capacity to ‘cope’ with emotional or stress changes throughout the day may be a more practical way to assist people with behavioral maintenance and weight control. This can be done in three ways, the first of which is equipping the individual with a wider set of strategies. There is evidence that the more resources a person possesses, the more resources they are likely to accrue [13]. A second strategy would be through improving a person’s psychological well-being. Positive well-being and optimism can improve resilience and the ability to problem-solve [14,15], as well as helping to restore resources after depletion [16]. Finally, a greater feeling of control over coping could equip a person with more confidence to use strategies. The Health Action Process Approach (HAPA) [17] suggests that self-efficacy (confidence) to initiate and maintain behaviors is critical to behavior change as people transition from action planning to coping planning. In other words, once the plan to lose weight has been initiated, individuals need to move on toward more coping-oriented planning in order to maintain healthy behaviors.

Thus, a supportive mobile phone app can be used to provide basic behavioral therapy with the intention of equipping people with greater self-awareness and better coping strategies, which, according to theory, could be associated with long-term behavior change. In some recent studies, a weight loss maintenance intervention using intelligent technology to assist people with diabetes to monitor their weight reported that higher use was associated with better weight loss results [18]. In a pilot trial, a behaviorally based iPhone app significantly improved psychological outcomes (mood and motivation) on a 2-month weight loss program [19]. However, no existing program grounded in behavioral theory has incorporated simple weight loss maintenance strategies into a supportive program that also targets well-being.

This paper will describe the development of a mobile phone app designed to use behavioral theory to target weight maintenance. By describing each of the core components and how they are intended to function, we aim to provide empirical and theoretical rationales for the inclusion of each of the app components as well as guidance for other developers regarding a potential approach for the development of app-based behavior change interventions.

Methods

Key Features of a Behaviorally Based Mobile Phone App for Weight Maintenance

We have attempted to design an app that aims to improve a user’s personal coping resources through basic behavioral therapy techniques that encourage workshopping of resources to deal with different moods and stresses while also providing weight, diet and exercise monitoring and utilizing immediate two-way feedback. Key features of the MotiMate app along

with the description of how they fit with theory and/or existing scientific evidence are described briefly below.

Behavioral Prompts

Simple SMS prompts can be useful for short-term behavior change [20]. There is also strong evidence to support the benefits of prompting and automated reminders to improve engagement with an intervention [21]. MotiMate prompts participants through push notification messages to enter data daily and review feedback weekly.

Monitoring and Reviewing

Self-monitoring is a key behavior change technique associated with successful behavior change [22] and weight control [23]. Setting and reviewing goals are core features of most behavioral treatments which would not be possible without a level of behavioral monitoring.

Monitoring tools were designed to encourage low intensity (<10 seconds) and frequent interactions, as can be seen in Figure 1A-D, are at the forefront of the user experience as 'homepage' of the app.

Weight Monitoring

Multiple studies have reported on the benefits of weight monitoring for weight control [24,25]. Real-time two-way feedback is displayed below the weight data as it is entered. This feedback is tailored based on existing definitions of weight maintenance: maintaining, danger zone and gaining [26]. Classification into one of these categories determines the nature of the textual feedback that people are given as well as the color of the weight display which, as can be seen in Figure 1A, progresses from green (maintaining) to dark green (danger zone) to grey (gaining). As participants enter the danger zone, the short automated message contains content designed to motivate them to stay on track. If they enter a gaining zone, the tone of these messages changes, remaining encouraging but becoming more directive, and an email is also sent to inform the administrator of the possible need for further intervention. Examples of messages for maintaining, danger zone and gaining messages respectively are: "Well done! You have maintained your starting weight. Keep it up," "You are a little heavier than your starting weight. Just a bit more effort this week and you will be back on track," and "You've gained some weight, try reviewing your lifestyle habits and keep keeping track of your weight."

Well-Being

Well-being monitoring was included to allow people to track their moods, so that they can see any link between emotional states and other behaviors (in summary sections) but also to allow a point of EMI for the coping behavioral support tool. Previous apps have tried to capture this style of processes successfully without persuasive features [27]. Mood states included were designed to capture pleasant and unpleasant moods as well as moods with high versus low arousal [28]. A user also identifies emotional intensity from 1-10 and location and time to allow review of data and identification of potential patterns in mood and to assist in the classification of coping thoughts and actions [29]. Stress (captured independently to mood) is measured through an intensity bar. No feedback text is presented below mood entries. The system detects and alerts an administrator via email if participants appear at psychological risk. The administrator's email address is unique to the app and monitored by the project manager for the trial version of the app. This person directs alerts to the appropriately qualified staff within the research institution (nutrition for weight gain and psychology for mood changes). Psychological risk is calculated by summing the intensity scores for each mood entered each day (with pleasant and unpleasant moods associated with positive and negative scores respectively). A summed negative score is used to indicate an "unhappy" mood day. The system sends an alert for a severely unhappy day (a summed score 3 standard deviations from the mean) or prolonged "unhappy" days (7 consecutive days).

Diet and Exercise Monitoring

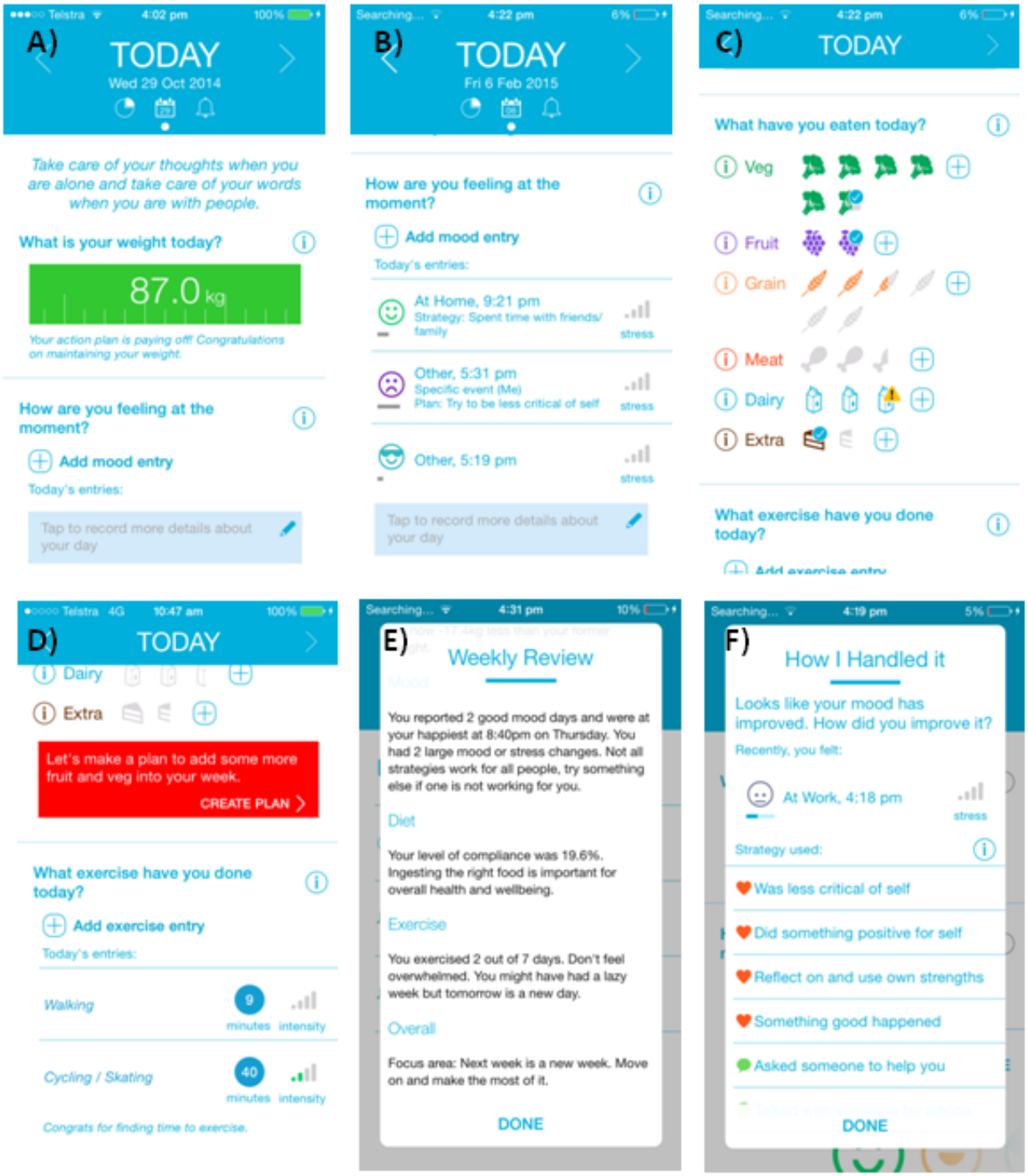
Given consistent evidence about the link between diet, exercise and weight loss maintenance [2], the app includes tools to monitor these behaviors. The aim is to design an approach that is generic enough to support a variety of different approaches for diet and exercise regimes that individuals had adopted in their weight loss journey. Therefore, national recommendations were used to provide feedback and set targets [30,31]. On creating an account, users are provided with personalized daily targets for each food group (e.g., fruit, vegetables, meat and dairy foods), displayed as grayed-out icons. If intake exceeds the target for a day, then an exclamation symbol appears for all food groups except fruit and vegetables. As can be seen in Figure 1C, to positively reinforce good eating habits, a tick appears if the user meets their target for all food groups (except for the junk food category, as less is always better). Figure 1D shows how automatic textual feedback is given on exercise records based on a series of rules formed using the Australian National Physical Activity Guidelines for adults and how many entries the user had made for the day.

In order to help keep users on track with their diet, an action planning interface is embedded in the food intake monitoring interface. When food intake consistently exceeds or falls below recommendations, users are provided with a predefined list of actions to choose from to help them "get back on track" and meet their food group targets.

Reviewing Data

Data from monitoring tools are collated into summary graphs to allow the users to reflect on their behaviors and identify potential patterns, as illustrated in Figure 1E. Once a week a behavioral review is also released that summarizes the user's weekly performance and gives feedback on which category a user is doing well on as well as their "focus" area (i.e. an area where they are performing poorly). Constructive reviews are critical in many behavioral therapies.

Figure 1. Screenshots of the MotiMate smartphone app. (A) opening page including motivational statement and weight entry; (B) Mood entries with description of hassles and coping strategies; (C) Food entries with examples of meeting and exceeding targets; (D) Exercise entry summary with prompt for diet action plan; (E) Weekly review (behavioural review); (F) Example of coping workshopping for improved mood.



Behavioral Support and Intervention

Supportive features in the app include motivating messages, personalized feedback in monitoring tools (as described above), and the coping tool. Previous studies have reported that supportive features can be beneficial for mood amongst women losing weight [19]. While some factors (eg, goal setting, motivation, and self-efficacy) may promote adherence to weight loss programs, psychological factors, such as stress and depression may inhibit peoples' ability to maintain weight losses [32-34]. Daily motivational messages were constructed based on inspirational quotes and thoughts to inspire optimism and the potential for higher resilience and problem-solving ability [14]. In accordance with HAPA theory, and as Figure 1A shows, these messages transition from a focus on general motivation and action planning through to coping planning.

Coping Behavioral Therapy Tool

Psycho-education contained within the information buttons provides guidance on maintaining positive mental well-being based on positive psychology and cognitive therapy. This component is designed to promote self-awareness and equip

people with greater self-efficacy to recognize and modify their behaviors in order to maintain their positive behavior changes.

The coping workshopping interface only appears once a large change in mood or stress is detected from the data entered. In order to capture information about both potential triggers and coping resources, the interface appears for both negative changes in mood or stress (eg, a decrease in happiness of 2 standard deviations or greater based on the group mean; or a good to a bad mood) and when mood or stress levels improve (eg, increase in happiness or a bad mood to good mood). Focusing on the coping resources that users have successfully applied rather than simply how they could fix hassles is an important behavioral element in the effort to assist users in building effective coping strategies.

Once the hassle interface is triggered, participants enter data on the nature of the issue, to whom it relates, and select a coping strategy they could use to address the issue. A range of coping strategies are presented in a predefined order using an algorithm assessing user appraisals of the hassle, including the immediacy of the issue, a user's perceived feeling of control and whether or not they want to fix the issue. Strategies are grouped generally into categories: social support (eg, "Talk with someone for advice"), personal strengths (eg, "Reflect on and use own strengths"), distraction (eg, "Avoid the issue") and emotional or cognitive strategies (eg, "Change my thinking about it") versus active strategies (eg, "Make a plan"). Escape/avoidant strategies could be associated with negative psychological symptoms [35] and therefore always appear at the bottom of this menu. When a positive change in stress or mood is entered, as Figure 1F shows, participants are asked what strategy they used to change their mood. In all menus, the option to add free-text to indicate something else is available. Once a person has workshopped their coping strategies, Figure 1B shows how the data are summarized on their home screen once a person has workshopped their coping strategies. Terms such as strategies and 'how I handled it' are presented to users in an effort to avoid negative pre-conceptions related to 'coping' [36].

Results

The development and trial of MotiMate received funding support in May 2014. All 88 volunteers started the trial by December 2014 and were in the process of completing their final visits when this paper was submitted (May 2015). Data analysis is currently underway and the first results are expected to be published early in 2016.

Discussion

Principal Findings

The paper has presented a scientifically informed mobile phone app to support weight loss maintenance. Many papers exist that propose or discuss behavioral theory and how this could be used to develop behavior change systems. Putting this into practice is challenging as it is often difficult to incorporate the number of features and depth required in such an evidence-based approach without compromising user interaction. We have used theory to guide the focus and ethos of MotiMate while also incorporating core behavioral therapy techniques, such as monitoring and reviewing. This has required drawing on insights from multiple theories (i.e., Conservation of Resources Theory) and models (i.e., HAPA) as well as scientific evidence from the literature. Despite this depth, we have attempted to keep user interactions simple by including multiple persuasive features. In an effort to facilitate shorter, more frequent interactions, the app does not collect detailed data. This means reliance on predetermined options and categories and a minimum level of user literacy. At this stage we have used the literature to guide the development of categories and provided "other" options where possible, but as more user data is collected, these categories will become more intuitive. More icons and graphical feedback may need to be incorporated into future versions to reduce reliance on high literacy levels for the two-way feedback.

Future Work

The MotiMate app is currently being trialed as part of a Randomized Control Trial to assess its efficacy (ANZCTRN 12614000474651). The trial requires participants to visit a clinic on 5 occasions over 24 weeks to have their weights taken and complete several psychological questionnaires. Data from this trial will help us to assess its efficacy as a persuasive tool for maintaining positive psychological well-being and weight relative to less supportive app. Detailed user feedback for each of the core features will be captured at weeks 4 and 12 to allow us to understand the relative strengths and weaknesses of each of the features. Usage logs of app features and correlation to behavior change are also being recorded throughout the trial. This will allow us to understand engagement over time relative to a basic monitoring app. It will also facilitate evaluation of how app use may change relative to changes in psychological well-being (such as increases in stress or depression) that will be collected during visits to the clinic. In a real-world application (outside the research institution), the alert system will require an administrator who can refer users to local primary care health services. The potential ongoing workload associated with this will also be evaluated to ensure its feasibility. In summary, the controlled trial will be a powerful method to allow assessment of the efficacy of the app for behavior change, capture app usage patterns, receive detailed user evaluation feedback, and assess feasibility of commercially releasing the app.

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Conflicts of Interest

None declared.

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Abbreviations

EMI: ecological momentary intervention
HAPA: Health Action Process Approach
SMS: short message service

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Original Paper

Assessing the Usability of Web-Based Alcohol Education for Older Adults: A Feasibility Study

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Abstract

Background: Older adults can experience unfavorable health effects from drinking at relatively low consumption levels because of age-related physiological changes and alcohol's potentially adverse interactions with declining health, increased medication-use and diminishing functional status. At the same time, alcohol use in older adults may be protective against heart disease, stroke, and other disorders associated with aging. We developed "A Toast to Health in Later Life! Wise Drinking as We Age," a web-based educational intervention to teach older adults to balance drinking risks and benefits.

Objective: To examine the intervention's feasibility in a sample of community-dwelling current drinkers ≥55 years of age and examine its effects on their quantity and frequency of alcohol use, adherence to standard drinking guidelines, and alcohol-related risks.

Methods: Participants were recruited in person, by mail and by telephone between September and October 2014 from a community-based social services organization serving Los Angeles County. Once enrolled, participants were randomly assigned to the intervention or to a control group. The conceptual frameworks for the intervention were the Health Belief Model, models of adult learning, and the US Department of Health and Human Services guidelines for designing easy-to-use websites. The intervention's content focuses on the relationship between drinking and its effects on older adults' medical conditions, use of medications, and ability to perform daily activities. It also addresses quantity and frequency of alcohol use, drinking and driving and binge drinking. The control group did not receive any special intervention. Data on alcohol use and risks for both groups came from the online version of the Alcohol-Related Problems Survey and were collected at baseline and four weeks later. Data on usability were collected online from the intervention group immediately after it completed its review of the website.

Results: The 49 intervention and 47 control participants did not differ at baseline in age, ethnicity, medication use, medical conditions, or alcohol use and both groups were mostly female, college-educated, and in good health. Of the intervention participants, 94% (46/49) had little or no difficulty using the website, with 67% (33/49) reporting that they will change the way they think about drinking because of their exposure to the education. At the 4-week follow-up, the intervention group reported drinking less (P=.02). No changes between groups were found in quantity and frequency, adherence to recommended guidelines, or risk status.

Conclusions: Community-dwelling older adults are receptive to online alcohol education. To be most effective, the education should be included as a component of a larger effort consisting of screening and counseling preferably in a health care setting.

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KEYWORDS

alcohol; older adults; elderly; web-based; education; online alcohol education

Introduction

Over 40% of US adults 65 to 74 years of age and 30% of adults 75 years and older are current drinkers [1,2]. Although the quantity and frequency of alcohol use tends to decline with age [1,3] consumption appears to be declining more slowly than in previous generations [4]. Older adults can experience unfavorable health effects even at relatively low consumption levels because of age-related physiological changes [5,6] and alcohol's potentially adverse interactions with chronic illness, increased medication-use and diminishing functional status [2-3,6,7].

Alcohol is implicated in many medical problems common in older adults including hypertension [8-11], depression [12,13], breast cancer [14-16], and fractures [17,18]. More than 60% of older adults regularly use medications [19], many of which, such as nonsteroidal anti-inflammatory drugs (NSAIDs), anticoagulants, and sedatives, have the potential to interact adversely with alcohol [7,20-22]. In addition, baby-boomers may have patterns of substance use that differ from previous cohorts resulting in a greater likelihood of combined alcohol and recreational drug use leading to increased risk for emergency department use and hospitalization [23-25]. Binge drinking (more than 4 or 5 drinks at one sitting), which greatly increases the chances of injury to self or others due to car crashes, violence, and suicide [26], is a serious problem in older adults who drink less intensely per occasion than their younger counterparts but binge more frequently than any other age group [26].

About 14.5% of older adults drink in excess of the National Institute on Alcohol Abuse and Alcoholism's (NIAAA) recommended limits [27], placing them at risk for alcohol-related problems. When health and drinking patterns are taken into account, about half of all drinkers 65 years of age or older may be at risk for experiencing alcohol-related harm even if they drink within recommended limits [2]. Considering that the number of older adults will increase to more than 20% of the US population by 2030 [28], the number of older people with alcohol-related risks and problems will increase even if drinking prevalence remains constant.

Accurate evaluations of alcohol-related problems in older persons are somewhat complicated by evidence that moderate consumption may have beneficial effects on cardiovascular functioning, stroke-prevention and all-cause mortality [29-36]. When compared to abstention, light to moderate drinking, such as 1 to 6 drinks weekly, is associated with a lower risk of incident dementia [37], congestive heart failure [38], rheumatoid arthritis [39], and diabetes [40,41].

Research and education tend to focus on alcohol use disorders, especially among younger people, and older adults usually do not have direct access to pertinent instruction that emphasizes drinking's potential benefits and risks that are specific to aging. Nor are data available on whether online alcohol education is usable by and acceptable to older adults, or if it can influence their drinking risks.

In response to the need for age-appropriate alcohol education, we developed and evaluated the feasibility of "A Toast to Health in Later Life! Wise Drinking as We Age," a Web-based program to educate older adults about the risks and potential benefits of drinking associated with aging. The Internet has been recognized for many years as an important mechanism for supplementing direct medical care [42-45]. Older adults are active Web users, with nearly 80% of persons 55 to 64 years and 60% of those 65 and older going online at least once a day [46]. Over two-thirds of seniors in their 70s are Internet users, and more than half use broadband [47].

People use the Internet because of several favorable features: the convenience of being able to search quickly for information at any time, unlimited access to inexpensive information, self-pacing, and user anonymity when searching for subject-sensitive health information [48-52]. This study evaluated "A Toast to Health in Later Life's" feasibility and influence on alcohol use and risks for alcohol-related problems in a sample of older current drinkers living independently in the community.

Methods

Participants and Setting

Participants were eligible for this study if they (1) were 55 years of age or older; (2) had 1 or more drinks containing alcohol in the past 3 months; (3) had an email account and were willing to share their email address so that staff could provide links to the educational program and study assessments; (4) were comfortable using the Internet; (5) had access to high-speed Internet; and (6) were willing to spend about 10 to 30 minutes on 2 separate occasions 4 weeks apart to complete an online alcohol education program and answer online questions in English about alcohol and health. Participants were recruited between September and October 2014 at a non-profit community-based social services organization located in Santa Monica, California and serving Los Angeles County. A trained study site coordinator used a combination of methods to recruit participants. These included in-person contacts with potential participants, mailings, and phone messages to the organization's database of volunteers and users. The organization's Chief Executive Officer also emailed qualified staff and colleagues with information about the study and requested their participation. Interested persons responded directly to the study's site coordinator and the CEO was not told who responded.

Study Design

The study site coordinator screened interested participants for their eligibility using a standardized script. Using a random number generator [53], we produced 200 numbers in random order. These numbers were allocated to each eligible study participant in the sequential order that they were generated. Participants with odd numbers were assigned to the intervention group, and those with even numbers were allocated to the control group. For example, the first 4 randomly generated numbers were 136, 172, 187, and 61. This meant that the first 2 eligible participants were assigned to the control group, whereas the next 2 participants were assigned to receive "A Toast to Health in Later Life!"

Participants in the intervention group received the URL for "A Toast to Health in Later Life!", a study user name, and a password. Once in the site, intervention participants were given the choice of watching a video or reading a transcript that explained the study's purposes and methods. If still interested, they clicked "submit" and were automatically directed to the informed consent form. After consenting, participants were automatically directed to the baseline survey, which collected information on drinking, health, medication use, functional status, lifestyle, and demographics. Participants were sent back to "A Toast to Health in Later Life!" automatically upon survey completion.

We programmed the website so that participants were required to review the entire site. Once they completed the review, however, participants were able to go back to any section that interested them. Intervention participants were given a brief post-survey focusing on the site's usability.

Control group participants completed the same online informed consent and baseline survey as the intervention group. They were offered access to the site after the completion of data collection.

The study's site coordinator emailed a link to the follow-up survey to all participants 4 weeks after each completed a baseline survey. Intervention and control participants completed identical follow-up surveys. Participants were reimbursed $35 after they completed all required data collection activities.

Conceptual Frameworks

The conceptual frameworks for this study and for use in guiding website content, questionnaire development, data collection and data analysis were The Health Belief Model, models of adult learning, and the US Department of Health and Human Services (DHSS) guidelines for designing easy-to-use websites [54].

We selected The Health Belief Model because it provides a framework for specifying personal and situational factors likely to influence health behavior [55], and it is among the most rigorously studied and commonly used models to study how education changes behavior [55-59]. According to the model, changing behavior is contingent upon several key factors including knowledge of the problem and its consequences and self-efficacy to do something about the problem. Throughout the website, we included facts ("Did You Know?") and practice exercises to reinforce learning. To strengthen self-efficacy, the site provides separate pages on how to cut down on drinking, how to speak to a doctor or other health professional about alcohol, and where to go for help. We used selected behavior change techniques, such as instruction on how to perform a behavior and demonstration of the behavior [60,61] to reinforce knowledge and self-efficacy.

To encourage learning, we relied on theories of adult learning which are often derived from or based on Knowles' theory, which he called "andragogy" [62,63]. Knowles' theory distinguishes adult from other learning in its emphasis on active learning, self-pacing, and problem-centered concepts [64,65]. The Internet is well suited to active learning and self-pacing. To facilitate active learning, "A Toast to Health in Later Life!" includes practice exercises and feedback.

We used DHSS guidelines [54] for easy-to-use websites (eg, keeping navigation simple and consistent; minimizing scrolling) to guide the website's development and separated the graphics arts component of Web development from the technical one so as to ensure that the site's graphics were colorful and age appropriate regardless of any technical challenge. The 2 study team geriatricians (JCB and DO), who have expertise in educating older learners, independently reviewed the website twice to make certain that it conformed to DHSS' usability criteria.

The Intervention

Content

"A Toast to Health in Later Life! Wise Drinking as We Age" is a Web-based education program that aims to teach older adults how to balance the benefits and risks of drinking. The website was developed by the study's team of alcohol researchers (AF and JCB), experts in geriatric medicine and education (JCB and DO), a computer programming expert (GY), and a Web designer (MR).

The website's intellectual content is derived from work done in connection with The Alcohol-Related Problems Survey (ARPS). The ARPS is a screening and education system that provides older adults and their physicians with tailored feedback on their alcohol-related risks based on their responses to a survey of alcohol use and health [66-73]. The ARPS system includes an educational component, which was developed with the assistance of over 200 community-dwelling older adults and then tested for usability in primary care practice with more than100 participants [74,75]. The study's geriatricians (JCB and DO) adapted the ARPS' educational content to "A Toast to Health in Later Life!" and used their expert knowledge and clinical experience to ensure the timeliness and relevance of the subject matter across the aging spectrum.

"A Toast to Health in Later Life!'" contains 9 sections, each of which provides information to achieve specific instructional objectives based on the ARPS' framework and content. For example, one objective is "to describe the relationship between drinking and medical conditions, medication use, and functional status in older adults." This objective is addressed throughout the site, but is emphasized in Section 1 of the website, "Thinking about Drinking and Aging." Sample content includes a statement such as: "Alcohol affects the workings of common medicines like high blood pressure medicines, pain killers, and antihistamines. Alcohol can also worsen or cause health problems such as cancer, heart disease, and depression. Also, some older people have problems walking, sleeping, or remembering things. Alcohol complicates the care of these problems."

Figure 1 contains a screenshot showing a portion of the content for the instructional objective, "to compare nonhazardous (wise), hazardous (risky) and harmful drinking."

Figure 1. Screenshot from "A Toast to Health in Later Life! Wise Drinking as We Age". Content for one instructional objective.

The Control

The control group did not receive any special intervention. Comparable patient education for older adults was not available, and we did not have the resources to develop an alternative and test it for feasibility and comparability.

Website Pilot Test

We conducted a 3-phase pilot test of the website among 21 people. The pilot test's purpose was to produce a website that was appropriate in content and usability and ready for a more comprehensive feasibility test. Participants in all phases of the pilot test met the same eligibility criteria as participants in the main feasibility study. The study's site yielded 17 participants, and 4 were recruited through personal contact. In the first phase of the pilot, we tested a prototype of the website on 5 older adults because evidence suggests that the best results come from testing no more than 5 users, and running as many small tests as you can [76]. The test asked participants to review the site without assistance. The study team (AF and JCB) then interviewed them in person or on the phone about the site's usability, appropriateness, and potential benefit. We revised the site based on participants' advice and retested it on 5 additional people who stated that they found the site easy to navigate, they learned from it, and they were confident that they could answer questions about alcohol use and aging. In the third phase of the pilot test, 4 people reviewed the website and answered questions about usability, while an additional 7 people reviewed the website and completed all study instruments.

Access to the Site

Access to the website required a user name (email address) and password (a-zA-z0-9) that was provided by the System Administrator. The System Administrator could grant System Administrator Privileges to others.

"A Toast to Health in Later Life!" was developed in Microsoft Razor MVC3 running under IIS. It is a dynamic website that presents information to users based on their progress through the site and their security profile. Beginning and end study surveys were hosted by Survey Monkey and embedded in the site's pages. The surveys were accessible to both the intervention and control groups, although the control group was not able to access any of the content pages.

Outcomes and Measures

Demographics

We used standard questions to ask participants at baseline about their sex, race and ethnicity, birthdate, and education. Participants were asked to rate their health status as being excellent, very good, good, fair, or poor.

Alcohol Use and Risks

To measure their alcohol-related use and risks, all participants completed the ARPS at baseline and 4 weeks after enrollment into the study. Since its development in the 1990's, the ARPS and its derivatives have been used with thousands of older adults in community and research settings to study alcohol's use and risks [2,67,77,78]. The ARPS' development, psychometric

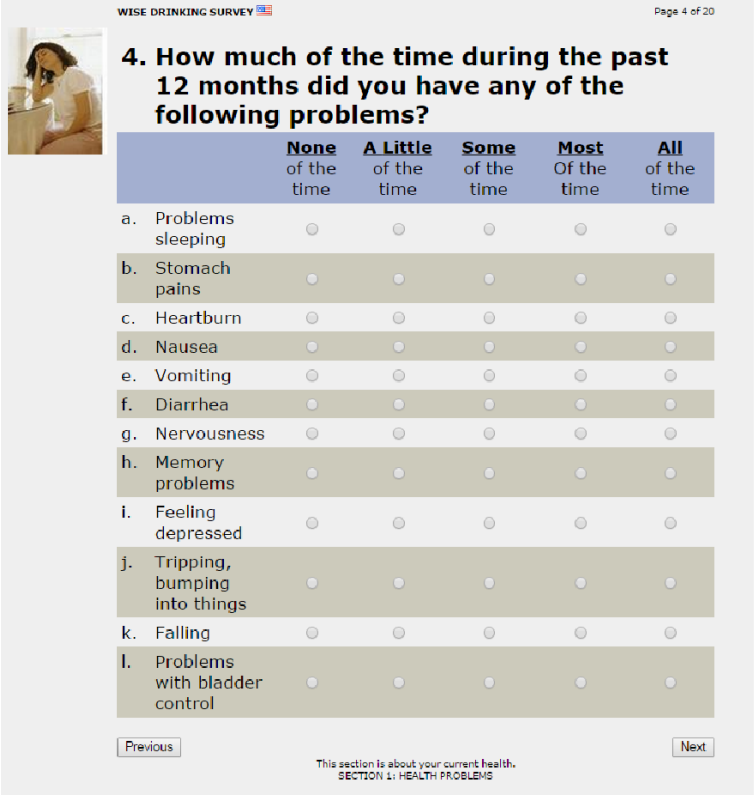
properties, and research use are well-documented [2,67,68,70,71,79]. A sample question from the ARPS is given in Figure 2.

The ARPS uses terminology [80,81] to classify alcohol-related health risk into 3 categories: harmful (consumption that may exacerbate or complicate existing alcohol-related problems), hazardous (consumption that poses risks of future harm for individuals with specific medical conditions, functional status, or symptoms, taking specific medications, or engaging in risky behaviors such as smoking), and nonhazardous (neither harmful nor hazardous and potentially beneficial).

The ARPS' scoring algorithms have been updated with the system's continued use in the United States and in other countries [82,83]. The algorithms first consider a person's reported quantity and frequency of consumption in relation to each of 63 factors (eg, medication use, binge drinking). The specific consumption patterns that confer risk vary substantially depending on the factor being considered. A sample of the tables and specifications for scoring the ARPS can be found in Multimedia Appendix 1.

To determine if NIAAA recommendations for older drinkers were exceeded [27], women and men 65 years of age and older who reported drinking 7 or more drinks weekly were considered at risk. All men under 65 years who drank 14 or more weekly drinks were also considered at risk.

Figure 2. Sample question from the Alcohol-Related Problems Survey (ARPS).



Usability

The intervention participants were automatically directed to a usability survey immediately upon completing their website review. The survey asked if participants learned about alcohol and health; if they were confident they could now answer questions about alcohol and health; and if they were confident, whether the website contributed to their confidence. We also asked participants if the website changed the way they now think about drinking, if they had any difficulty using the website and if they would recommend it to others.

Ethics and Informed Consent

Eligible participants were asked to complete online consent after they were given the opportunity to listen to and read an overview of the study's purposes and procedures. The informed consent took the form of an information sheet with the following categories: the study's purposes and methods, a description of participant expectations, a discussion of possible risks or discomforts associated with participation, a discussion of possible benefits, payment for participation, measures used to protect privacy, whom to contact if questions arose, rights as a research subject, and participants' right to quit the study at any time. Participants who clicked on submit after reading information sheet were automatically directed to the study's baseline survey. Participants were free to close their browsers at any time, thus leaving the study, but intervention participants who completed the baseline survey and also agreed to continue, clicked on a link that automatically directed them to the study's website. Entry to all study materials was protected by password and all responses were collected over secured, encrypted SSL/TLS connections. An independent Institutional Review Board (US Office of Human Subjects Research IRB #0000667) approved this study. A copy of the informed consent form can be found in Multimedia Appendix 2.

Data Analysis

Analyses of outcome data were based on the per-protocol method which is commonly used in feasibility studies because of their exploratory nature. Thus, data were analyzed for the 47 control and 49 intervention group participants who provided complete data on all outcome measures.

The outcomes were measured 4 weeks after enrollment and included (1) the quantity and frequency of drinking as assessed by the ARPS; (2) the percentage of participants drinking above NIAAA recommended levels; (3) the percentage of participants who were harmful, hazardous and nonhazardous drinkers as assessed by the ARPS; and (4) whether participants report changing their drinking amount in the past 4 weeks as indicated by their answers to survey questions.

Baseline characteristics are reported for the intervention and control groups. Categorical data are reported as frequencies (percentages), continuous data are reported as means and standard deviations (SD), and non-normally distributed data are reported as medians and ranges. We used Chi-square tests (or Fisher's exact tests) for categorical data and *t* tests for continuous data (or Wilcoxon-Mann-Whitney tests for non-normally distributed data) to analyze differences between the 2 groups. To determine any changes from baseline to follow-up, we used Chi-square tests (or Fisher's exact tests) to test group distributions at baseline versus follow-up. As a sensitivity analysis, we excluded participants who were nonhazardous at baseline to compare changes among hazardous and harmful drinkers. We also ran paired data analyses with McNemar's test (or Bowker's test of symmetry as appropriate) to determine any changes from baseline to follow-up within the intervention and control groups.

All tests were 2-sided, and statistical significance was set at .05. All analyses were conducted with SAS 9.4 (SAS Institute).

Results

Study Participants

Over the study's 3-month recruitment period, we screened 137 interested participants (Figure 3), 20 of whom were not eligible for participation. There were no differences between control and intervention groups in enrollment or attrition. Of the 112 participants who were randomized, 91.0% (51/56) of the control and 89.0% (50/56) of the intervention group completed the baseline assessment. No differences were found in sources of recruitment for participants who completed the baseline assessment, with 72.5% (37/51) of the control and 72.0% (36/50) of the intervention having been recruited from the study site's volunteers. Four weeks after enrollment, 92.0% (47/51) of the control and 98.0% (49/50) of the intervention group completed all study measures.

The intervention and control groups did not differ in gender; age; education; ethnicity; or alcohol use at baseline, and they were mostly female; college educated; and in good health.

Table 1. Intervention and control participant baseline characteristics.

	Intervention Group (n=49) n (%)	Control Group (n=47) n (%)	*P*
Sex (Female)	34 (69.4)	33 (70.2)	.93
Education, % College or more	39 (79.6)	41 (87.2)	.33
Health status, % Excellent or very good	37 (75.5)	31 (66.0)	.30
Race: White	44 (89.8)	42 (89.4)	.99[a]
Ethnicity: Hispanic	3 (6.1)	3 (6.4)	
Doctor ever told you have hypertension	21 (42.9)	21 (44.7)	.86
Doctor ever told you have breast cancer (♀)	4 (11.8)	5 (15.2)	.74[a]
Doctor ever told you have depression	6 (12.2)	8 (17.0)	.51
Falls were a problem in the past 12 months	6 (12.5)	8 (17.0)	.53
Daily NSAIDs[c]	13 (26.5)	12 (25.5)	.91[a]
Daily high blood pressure medicine	20 (40.8)	17 (36.2)	.64
Daily take an anticoagulant	1 (2.1)	1 (2.2)	.99[a]
Weekly sedative, narcotic or tranquillizer	11 (22.5)	8 (17.0)	.50
Ever drank 4 or more drinks at one sitting in past 12 months	8 (16.3)	11 (23.4)	.38

[a] Fisher exact test

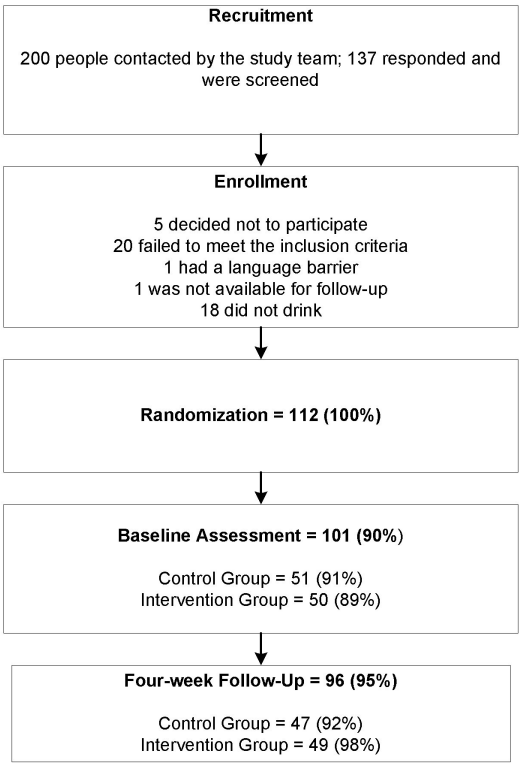
[b] Wilcoxon-Mann-Whitney test

[c] NSAID: Nonsteroidal anti-inflammatory drug

On average, participants in each group were 70 years of age and White. In the intervention group 42.9% (21/49) participants reported that their doctor told them they had hypertension and 40.8% (20/49) were taking antihypertensives. Similarly, 44.7%

(21/47) of the controls reported having been told they had hypertension, and 36.2% (12/47) were using antihypertensives. Of the 13 intervention participants 26.5% (13/49), and 12 controls 25.5% (12/47) used NSAID's daily. Weekly drinking varied widely, with members of the intervention group drinking from 0 to 28 drinks, and members of the control group drinking from 0 to 21 drinks. Of the intervention group, 16.3%) (8/49) of the intervention and 23.4% (11/47) of the control group participants drank 4 or more drinks at one sitting in the past 12 months.

Figure 3. Participant flow chart.



Drinking Quantity and Frequency

Drinking quantity did not change over time for the intervention (*P*=.31) or control group (*P*=.56) (Table 2).

At baseline, 89.8% (44/49) of the intervention and 97.8% (46/47) of the control group drank 2 drinks or less when they drank, and at follow-up, 96.0% (47/49) of the intervention and 93.6% (44/47) of the control group drank 2 drinks or less. Similarly, the frequency with which study participants drank did not change significantly from baseline to follow up for the intervention (*P*=.99) or the control group (*P*=.67). Paired data analyses yielded similar results for quantity and frequency of drinking (all *P*-values >.05).

Adherence to NIAAA Recommended Weekly Limits

The proportion of participants who exceeded NIAAA's recommended weekly limits did not significantly change over time in the intervention (*P*=.99) or control (*P*=.53) group (Table 3). At follow-up, 30.6% (15/49) of the intervention and 48.9% (23/47) of the control group's drinking exceeded the NIAAA's limits. No differences were found in the paired data analyses.

Table 2. Quantity and frequency of drinking.

Quantity/Frequency	Intervention	Control					
		Baseline (n=49) n (%)	Follow-up (n=49) n (%)	*P*	Baseline (n=47) n (%)	Follow-up (n=47) n (%)	*P*
Quantity							
	3 or more drinks per day[a]	5 (10.2)	2 (4.1%)	.31[b]	1 (2.1%)	3 (6.4%)	.56[bd]
	2 drinks per day	9 (18.4)	14 (28.6%)		16 (34.0%)	18 (38.3%)	
	1 drink or less	35 (71.4)	33 (67.4%)		30 (63.8%)	26 (55.3%)	
Frequency							
	Drinks daily or almost daily[c]	15 (30.6)	14 (28.6)	.99[b]	18 (38.3)	18 (38.3)	.67[d]
	Drinks 4 or 5 times a week	4 (8.2)	5 (10.2)		6 (12.8)	9 (19.2)	
	Drinks 2 or 3 times a week or less	30 (61.2)	30 (61.2)		23 (48.9)	20 (42.6)	

[a] No differences in quantity between the groups at baseline (*P*=.09) or follow-up (*P*=.54).

[b] Fisher's exact test

[c] No differences in frequency between groups at baseline (*P*=.46) or follow-up (*P*=.17).

[d]*P*-values for the paired data analyses (Bowker's test of symmetry) between baseline and follow-up for quantity is .26 for the intervention group and .26 for the control group; for frequency, the *P*-value is .80 for the intervention group and .36 for the control group.

Table 3. Participants who passed and failed to meet NIAAA's[a] recommended weekly limits.[b]

Result	Intervention			Control		
	Baseline(n=49) n (%)	Follow-up(n=49) n (%)	*P* (by time)	Baseline (n=47) n (%)	Follow-up (n=47) n (%)	*P* (by time)
NIAAA fail[c]	15 (30.6)	15 (30.6)	.99[d]	20 (42.6)	23 (48.9)	.53[d]
NIAA pass	34 (69.4)	34 (69.4)		27 (57.4)	24 (51.1)	

[a] NIAAA: The National Institute on Alcohol Abuse and Alcoholism

[b] 7 drinks weekly for all women and men 65 years of age and older; 14 drinks weekly for men under 65 years of age

[c] No differences between groups at baseline (*P*=.22) or follow-up (*P*=.67).

[d]*P*-values for the paired sample analyses (McNemar's test) between baseline and follow-up are .99 (100% agreement) for the intervention group and .26 for the control group.

Change in Drinking Risk

At baseline, 44.9% (22/50) of intervention and 66.7% (31/51) of control group were harmful or hazardous drinkers (Table 4). No changes occurred from baseline to follow-up for either group in any category. There were also no differences in the risk categories between intervention and control at either baseline or follow-up (*P*=0.12 and *P*=0.11, respectively). This remained true in the sensitivity analysis where we excluded the participants that were nonhazardous drinkers at baseline (*P*=0.94 at baseline and *P*=0.58 at follow-up) (data not shown).

Table 4. Drinking risk.

Risk level	Intervention			Control		
	Baseline[a] (n=49) n (%)	Follow-up[a] (n=49) n (%)	*P*	Baseline[a] (n=47) n (%)	Follow-up[a] (n=47) n (%)	*P*
Harmful[c]	13 (26.5)	16 (32.7)	.63	18 (38.3)	21 (44.7)	.74[b]
Hazardous[d]	9 (18.4)	6 (12.2)		13 (27.7)	10 (21.3)	
Non-hazardous[e]	27 (55.1)	27 (55.1)		16 (34.0)	16 (34.0)	

[a] No differences in risk category at baseline (*P*=.12) or follow-up (*P*=.11).

[b]*P*-values for the paired data analyses (Bowker's test of symmetry) between baseline and follow-up are .73 for the intervention and .72 for the control group.

[c] Problems are likely

[d] At-risk for problems

[e] No known risks

Change in Drinking From Baseline to Follow-up

Participants were asked if they changed their drinking in the past 4 weeks, and if so, whether they drank more, the same, or less. Table 5 shows that although intervention group did not differ from control group at baseline (*P*=.74), the intervention group differed significantly from the control group at follow-up (*P*=.02), with intervention participants reporting that they drank less.

Table 5. Participants reported change in drinking in the past 4 weeks.

	Baseline		*P*	Follow-up		*P*
	Baseline[a] (n=49) n (%)	Follow-up[a] (n=47) n (%)	.74	Baseline[a] (n=49) n (%)	Follow-up[a] (n=47) n (%)	.02[b]
Less	6 (12.2)	4 (8.5)		13 (26.3)	4 (8.5)	
Same	0 (0.0)	0 (0.0)		0 (0.0)	0 (0.0)	
More	43 (87.8)	36 (73.5)		37 (73.5)	43 (91.5)	

[a]The Fisher exact test *P* value for the control group from baseline to follow-up is .99.

[b]*P* value for the paired data analyses (McNemar's test or Bowker's test of symmetry) is .02 for the intervention group from baseline to follow-up; for the control group, the *P*-value is .99.

Usability

On average, the 49 intervention participants spent between 10 and 20 minutes on the website. The majority (93.9%, 46/49) reported having little or no difficulty using the site (Table 6). Most also reported (96.0%, 47/49) reported that they learned from the website, and 75.5% (37/49) stated being very or somewhat confident that they can answer questions about alcohol and health. A large proportion (77.6%, 38/49) indicated that the website helped them to become more confident in their ability to answer questions, and 93.9% (46/49) definitely or probably would recommend the site to others. Finally, 67.3% (33/49) said the use of the site will change the way they think about drinking.

Table 6. Intervention participants' report on the usability of "A Toast to Health in Later Life!" (n=49).

Usability characteristic		n (%)
Difficulty using the site		
	Rated "No difficulty"	36 (73.5)
	Rated "Little difficulty"	10 (20.4)
Learned from the site		
	Rated "A great deal"	32 (65.3)
	Rated "A little"	15 (30.6)
Confident can answer questions about alcohol		
	Rated "Very"	12 (24.5)
	Rated "Somewhat"	25 (51.0)
Website increased confidence		
	Rated "Definitely"	18 (36.7)
	Rated "Probably"	20 (40.8)
Recommend to others		
	Rated "Definitely"	30 (61.2)
	Rated "Probably"	16 (32.7)
Change the way you think about drinking		
	Rated "Some"	32 (65.3)
	Rated "Much"	1 (2.0)

Discussion

Principal Results

We developed and tested the feasibility of a "Toast to Health in Later Life!" with older adults who were comfortable using the Internet and lived independently within the community. Most intervention participants had little or no difficulty navigating the website and recommended it to others. The participants reported that they learned from the site, could now confidently answer questions about alcohol and aging, and agreed that the information they gathered will change the way they think about drinking. Intervention participants reported drinking significantly less than controls 4 weeks after study enrollment.

Despite the intervention group's favorable review of the website, and their perception that they drank less because of it, we found no difference between the intervention and control participants in their quantity and frequency of drinking, adherence to NIAAA weekly drinking guidelines, or drinking risks over the study's 4-week data collection period.

There are several explanations for the discrepancy between participants' favorable perceptions and the study's statistical findings. First, notwithstanding the site's appeal to participants, the study may not have provided the supportive resources necessary for them to change their drinking behavior. Other successful efforts to reduce hazardous and harmful drinking in older adults have provided access to screening, personalized feedback, health care provider advice and other services in addition to education. In Project GOAL [84], one of the first studies to target older adults, physicians used a work-book to give advice to patients, included a second reinforcement visit with the physician, and provided 2 follow-up calls from a clinic nurse 2 weeks after each physician visit. These features probably contributed to its success in reducing both 7-day alcohol use and binge drinking.

Using the ARPS screening and education system, a study of 665 older adults and 23 physicians in the same community as the present study provided screening and personalized feedback to patients and their physicians in addition to patient education. Patients significantly reduced harmful drinking at follow-up from a statistically expected 21% in usual care to 16% and increased nonhazardous drinking from 52% expected in usual care to 58% over a 12-month period [67].

Another study of 1186 community-dwelling older drinkers and 31 primary care providers [77] succeeded in reducing at-risk drinking as defined by a measure based on the ARPS. In addition to providing participants with educational materials, this study also provided participants with personalized reports, drinking diaries, physician advice during office visits, and telephone counseling delivered by a health educator. Similarly, a study [78] which included 631 older at-risk drinkers, also using measures derived from the ARPS algorithms, found that its multi-faceted intervention among older at-risk drinkers in primary care did not reduce the proportions of at-risk or heavy drinkers but the intervention was found to reduce the amount of drinking at 12 months [77]. This study's participants were given personalized reports, a booklet on alcohol and aging, a drinking diary, and received advice from a primary care provider and telephone counseling from a health educator.

These studies took place in medical rather than community settings. The presence and support of health personnel is likely to have motivated participants, encouraging them to change their drinking behavior. The effective use of health personnel to motivate patients has been shown in clinical settings for problems such as preventive health care use, health behaviors, and control of diabetes and hyperlipidemia [85,86].

Another possible explanation of the present study's inability to detect differences in quantity and frequency and in risks is that its brief duration may have been insufficient for change to occur. Many participants may not have seen a health care provider over the study's 4-week data collection period, eliminating the potential for alcohol-related discussions on how to make changes in lifestyle or medication-use. Further, participants may not have encountered situations, such as a party or other festive occasion, in which to change their drinking pattern. Additionally, intervention participants may have reported drinking less because they perceived it to be the socially desirable or expected response.

Limitations

This study's findings must be interpreted cautiously. The study was an initial test of the website, and there were no comparable studies on which to base sample size calculations. Thus, we cannot be sure if the number of participants was sufficient to detect a true difference between groups. Further, we did not compare "A Toast to Health in Later Life's feasibility with alternative approaches or programs designed to educate older people about the benefits and risks of alcohol drinking because, to our knowledge, none was available. In fact, it was the lack of existing age-appropriate education that was the impetus for developing and testing "A Toast to Health in Later Life." We might have developed our own alternative version of the content (eg, audio or print), but that would have necessitated a systematic test of the feasibility and comparability of this new program (eg, for literacy level and age relevance of content), and we did not have the resources to conduct such a study. Instead we took the approach advocated by some researchers who have stated that even if evaluations find that one intervention is superior to an alternative intervention, they cannot claim that the superior treatment should be the new standard because the intervention has not been shown to be superior to the care commonly given by practitioners [87].

This study's sample included nonhazardous drinkers who may not have had an incentive to change their drinking patterns, raising questions about the appropriateness of including them in the study. We included these participants because previous research has shown that nonhazardous drinking older adults may become risky drinkers over time. In a study of community-dwelling older drinkers, for example, 20 of 112 (18%) of nonhazardous drinkers who did not receive an intervention became hazardous or harmful consumers over the study's 12-month period [67]. Also, because this study aimed to test the website's feasibility, we considered all current drinkers to be eligible, and the sensitivity analysis did not change the study's findings. Given that there is evidence that without intervention, some older drinkers may increase their risks, it seems prudent to educate all older drinkers regardless of their current risk. Future research should consider evaluating the effectiveness of patient education in preventing alcohol-related risks in older people as well as reducing those that already exist. Enthusiasm for the applicability of the study's findings must also be tempered by the fact that the participants were selected because they were comfortable using the Internet. In the United States, nearly 60% of persons 65 and older use the Internet, with persons 55 years and older using it even more frequently [47,88]. These statistics may not apply to other US samples or nations, thereby limiting the feasibility of "A Toast to Health in Later Life" among many older adults.

Conclusions

This study was designed to test "A Toast to Health in Later Life's!" feasibility and determine if education has the potential to change older adults' drinking behavior. We found that the site is usable and acceptable in a White, healthy, and well-educated sample of older people. We do not know if older adults with other demographic characteristics will react to the website in the same positive way. But in terms of its drinking, the study's sample was comparable to other older adults [2,67] in its frequency of drinking risks and failure to adhere to NIAAA drinking limits. This finding supports the suggestion that the website's objectives and content are appropriate for a more general US population.

Despite the study's limitations in setting, duration, and sample characteristics, it is the first that we know of to provide evidence that older adults are willing and able to use online education to learn about alcohol use and aging. Its findings also suggest that for alcohol education to be most effective, it should be included as a component of a larger effort consisting of screening, education and counseling preferably in a health care setting. In the United States, Medicare reimburses health providers who screen older patients and counsel those who are at-risk but do not meet the criteria for alcohol use disorders. Given how many preventive activities are expected of primary care providers [89], online education programs, such as "A Toast to Health in Later Life!" can be used to supplement counseling, possibly ameliorating the health care provider's burdens especially with respect to sensitive topics such as alcohol use. Important next steps consist of testing the usefulness of "A Toast to Health in Later Life!" or other Web-based alcohol education programs in larger studies on relatively diverse older populations and also evaluating their effectiveness and cost-effectiveness.

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Authors' Contributions

AF was responsible for the study's conceptualization and quality. She contributed significantly to the study's design, data collection, data analysis, and reporting. LK was responsible for the statistical analysis and writing the results and findings. DO was instrumental in conceptualizing the study and in reviewing the website's content and the study's findings. JVD supervised the randomization procedure and data collection and contributed to the writing. AC assisted in data collection and writing. JCB contributed to the study's conceptualization and quality and oversaw the production of the website and its intellectual quality. He also participated in data collection and in writing this manuscript.

Conflicts of Interest

Arlene Fink Associates has received SBIR grants in the area of Web-based alcohol interventions and owns the intellectual property of the application discussed. None of the authors except AF is employed by Arlene Fink Associates, holds equity or stock options or benefits in any other way from the publication of this article. AF owns 100% of Arlene Fink Associates and may benefit from publication of this article. JCB has access to all data and acts as independent guarantor for this study. Other authors have no conflicts of interest to declare.

Multimedia Appendix 1

A portion of the tables and specifications for the Alcohol-Related Problems Survey.

[PDF File (Adobe PDF File), 206KB - resprot_v5i1e11_app1.pdf]

Multimedia Appendix 2

Informed consent.

[PDF File (Adobe PDF File), 51KB - resprot_v5i1e11_app2.pdf]

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Abbreviations

ARPS: Alcohol-Related Problem Survey
DHSS: Department of Health and Human Services
NIAAA: National Institute of Alcohol Abuse and Alcoholism
NSAID: Nonsteroidal anti-inflammatory drug

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Original Paper

Evidence-Based mHealth Chronic Disease Mobile App Intervention Design: Development of a Framework

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Abstract

Background: Mobile technology offers new capabilities that can help to drive important aspects of chronic disease management at both an individual and population level, including the ability to deliver real-time interventions that can be connected to a health care team. A framework that supports both development and evaluation is needed to understand the aspects of mHealth that work for specific diseases, populations, and in the achievement of specific outcomes in real-world settings. This framework should incorporate design structure and process, which are important to translate clinical and behavioral evidence, user interface, experience design and technical capabilities into scalable, replicable, and evidence-based mobile health (mHealth) solutions to drive outcomes.

Objective: The purpose of this paper is to discuss the identification and development of an app intervention design framework, and its subsequent refinement through development of various types of mHealth apps for chronic disease.

Methods: The process of developing the framework was conducted between June 2012 and June 2014. Informed by clinical guidelines, standards of care, clinical practice recommendations, evidence-based research, best practices, and translated by subject matter experts, a framework for mobile app design was developed and the refinement of the framework across seven chronic disease states and three different product types is described.

Results: The result was the development of the Chronic Disease mHealth App Intervention Design Framework. This framework allowed for the integration of clinical and behavioral evidence for intervention and feature design. The application to different diseases and implementation models guided the design of mHealth solutions for varying levels of chronic disease management.

Conclusions: The framework and its design elements enable replicable product development for mHealth apps and may provide a foundation for the digital health industry to systematically expand mobile health interventions and validate their effectiveness across multiple implementation settings and chronic diseases.

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KEYWORDS

mHealth; mobile applications; mobile app design; chronic disease; diabetes; mHealth framework; behavioral intervention; intervention design; mHealth implementation; telemedicine

Introduction

The promise of mobile technology to revolutionize health care services and patient self-management behavior for chronic disease has intrigued practitioners and researchers for well over a decade [1]. The attributes that give it an advantage over other information and communication technologies are its popularity, mobility, and technological capabilities [2]. Mobile health (mHealth) interventions can benefit health care by reaching people in resource-poor settings, delivering interventions to large numbers of people, claiming people's attention when it is most relevant, enabling access to and delivery of customized support, and providing low cost interventions [3]. Mobile technology offers new capabilities that can help to drive important aspects of chronic disease management at both an

individual and population level, including the ability to deliver real-time interventions that can be connected to a health care team.

Researchers, technology companies, health care companies, health plans, and pharma have been exploring these capabilities across different chronic disease management programs and models**.** In order to design mHealth solutions, practitioners and researchers can draw from innovation, best practices, theory, and evidence. When building different mHealth products for a variety of settings and programs, the type of mHealth product developed is dictated by the objectives deemed necessary for the population being served. For example, building mHealth solutions that provide comprehensive management of chronic disease will differ from building those that support one component of chronic disease management in an existing health care program.

Over the past decade, the evidence base indicating the efficacy and effectiveness of these mHealth technology solutions has been growing for the management of chronic disease. However, evidence of their effectiveness has been inconclusive [3-5]. Some literature has provided preliminary evidence regarding the utility of linking mHealth into existing health care models to help drive improved outcomes [6-11]. mHealth apps have utilized many intervention strategies such as tracking and texting to offer more comprehensive management support [8,12]. The inconclusive findings highlight the importance of the following questions: which aspects of mHealth work for which diseases, and for whom, to achieve which outcomes?

In order to answer these questions, a systematic approach to designing mHealth apps to support health programs and services becomes vitally important. To date, there is very little to guide such a process [13], which facilitates translation of the emerging mHealth science and literature into scalable, replicable, evidence-based mHealth solutions that can be adapted to multiple, real-world health care settings and systematically evaluated. Through the experience of developing mHealth products for different health care settings, there was a unique opportunity to develop and test systematic approaches for designing and developing mHealth interventions. Informed by the Chronic Care Model [14], health behavior models and theories, clinical and behavioral program best practices [15-18], and health care outcomes, an app design framework evolved. The purpose of this paper is to discuss the identification and development of an app intervention design framework, and its subsequent refinement through development of various types of mHealth apps for chronic disease.

Methods

Phase 1: Developing the Initial Framework (June to August 2012)

As a first step in creating a systematic process for app intervention design, the Clinical Programs and Research subject matter experts (SME) identified the strategic, intervention, and program domains that were the foundation of a telephonic disease management program that had demonstrated positive outcomes [19]. In that program, patient interventions were delivered telephonically by a case manager. For mobile app intervention development, we needed to understand how to leverage the anywhere, anytime, contextual capabilities of mobile technology, as well as employ existing evidence and expertise to modify, adapt, and incorporate traditional interventions in the context of mobile app product design. In collaboration with the Behavior Change SME and other clinicians, the initial framework (strategic, intervention, and program domains) was translated into a framework that was specific to mobile app intervention design.

The strategic domains were expanded to include value drivers, outcomes and metrics, and program objectives as defined by key stakeholders. Since mobile health intervention development is still in an early stage, it was essential to understand what types of intervention(s) could be delivered through an app and how success would be measured. For example, tracking a metabolic measure such as blood pressure or weight required different features than providing tailored behavioral support that could impact clinical and behavioral outcomes.

Through the collaborative process, the intervention domain was expanded to integrate appropriate clinical and behavioral components that were informed by clinical guidelines, standards of care [20], clinical practice recommendations, evidence-based research, best practices, and programmatic expertise for chronic disease. The clinical, program, and behavioral experts reviewed all the clinical resources and leveraged existing programmatic guidelines [15-18,21] and experience for developing behavior change strategies. Three intervention domains were added: essential behaviors (supporting actions and determinants), multidimensional profiles, and evidence-based clinical and behavioral interventions. Together these domains make up the intervention plan. This plan then informs the design of the specific product features and content that support the intervention(s) and drive outcome(s).

Phase 2: Applying and Refining the Framework (September 2012 to October 2013)

After the initial development, the framework was applied to design three types of mHealth products (apps) providing a range of chronic disease support and management: (1) direct-to-consumer apps providing targeted intervention(s) such as tracking, reminders, and data display (eg, symptom tracker); (2) program apps developed for “single-focus programs” (eg, low back pain); and (3) prescription apps providing comprehensive chronic disease support through numerous features and capabilities linking the patient and health care team (eg, mobile prescription therapy). Initially, the framework was applied to type 2 diabetes and refined. With each application of the framework to a new disease state and product/program, domains were validated and components for each domain were refined, such that an initial taxonomy of app intervention design evolved.

Phase 3: Finalizing the Framework (March 2013 to June 2014)

Through collaboration with clinical informatics SMEs and software developers, and the application of the framework domains to the development of different types of mobile apps,

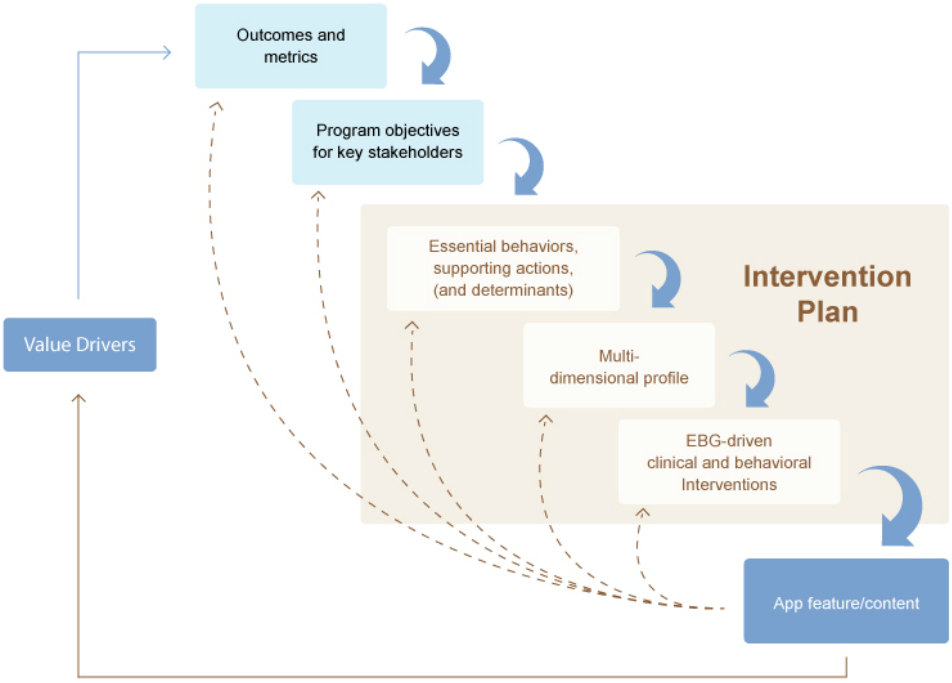
the finalized framework and taxonomy for an mHealth app intervention design evolved. The taxonomy reflected the framework domains and associated design elements for each domain attribute.

Results

The Design Framework

The Chronic Disease mHealth App Intervention Design Framework resulted from an iterative process in which the framework was applied, evaluated, and refined during the design and development of different types of apps for 7 chronic diseases. The framework includes 7 domains to guide the development of apps: 3 strategic, 3 intervention design, and 1 product feature(s) and content. As the work for each domain is completed, the output is fed into the next domain in what is described as a "waterfall process" (Figure 1). This process guides both the development of the product features (downward arrows) and the evaluation (dotted upward arrows) of their effectiveness in driving desired outcomes. Application of the framework in a systematic way results in the identification of key features and content to be included in mHealth products. The specific domains evolved because they provide a coherent means of explicitly describing the logic behind major decisions related to the features and content included in mHealth products. They also guide the measurement of the process, outcomes, and impact that the mHealth app brings to the product/program. Application of the framework results in apps that are either direct-to-consumer tracking apps, "single-program" apps, or mobile medical apps regulated by the Federal Drug Administration (FDA). Insights gained in relation to design decisions for different types of apps demonstrated the value of the framework. A comparative analysis of different apps will be presented in subsequent publications.

Figure 1. mHealth waterfall process.



The Framework

The following is a description of the framework and its 7 domains. For each domain, we present a definition and how the domain was derived. The first 3 domains (value drivers, outcomes, and program objectives) are strategic and were determined through interaction with external stakeholders.

Value Drivers

Definition

Value drivers are the entities that increase the value of a product or service [22]. Examples for an mHealth product can include improved health for an individual or population, improved access to care, or reduced health care costs.

How Derived

Value drivers were initially determined by analyzing gaps and challenges in chronic disease management at the population level, in service delivery, and in patient self-management. For each mHealth product, value drivers were informed by the overall goals and objectives of the program and by the unique capabilities that an mHealth app could contribute to the improvement of chronic disease management, service delivery, and/or patient self-management. It was also necessary to take into account the setting in which the mHealth solution would be deployed (eg, primary health care, large employer-based insurance program, large pharmaceutical). Value drivers set the course for the development of the mHealth solution, and in turn, dictated subsequent elements of the "waterfall."

Outcomes and Metrics

Definitions

Outcomes are the desired results of the program, and generally can be short (eg, knowledge, attitudes), intermediate (eg, self-care behaviors), or long term (eg, A1c) depending on the

objective, length of the program, and expectations of the program or intervention(s) [23]. Metrics are a means of measurement and are aligned to industry-recognized quality metrics to facilitate a program or providers' ability to demonstrate that intervention(s) have an impact.

Derivation

Guided by the designated value drivers for each program, outcomes and metrics were aligned with those of the chronic disease program being supported. National guidelines, standards of care, published literature, and meta-analyses were common sources for identifying standard outcomes and metrics for each product.

Program Goals for Key Stakeholders

Definition

Key stakeholders are those users that the system is designed to support directly. The program goals for key stakeholders represent what the intended users should accomplish via product use. Typically, they signify fundamental, long-range achievements based on engagement with the mHealth product and are broad general statements [24].

Derivation

Based on the value drivers, outcomes, and metrics, key stakeholders and corresponding program goals for those stakeholders were identified by interdisciplinary teams responsible for developing the mHealth programs and products.

Intervention Plan

The intervention domain was expanded to include 3 subdomains that resulted from the translation and application of current behavioral and clinical evidence, as well as subject matter expertise to the real-time, contextual capabilities of mobile technology: essential self-management behaviors, multidimensional profiles, and integrated clinical/behavioral interventions. These 3 domains were grouped together as the intervention plan.

These domains guided the design of integrated clinical and behavioral interventions that target behaviors known to improve clinical outcomes. Behavioral interventions were designed based on clinical contexts to guide intended value drivers and outcomes that uniquely address the chronic disease of interest. Several of the most commonly used behavior change theories guided/informed these domains [25]; specifically, determinants addressed through product design included the social ecologic model [26], the health belief model [27], social cognitive theory/social learning theory [28], theory of reasoned action and planned behavior [29], and the transtheoretical stages of change model [30]. Disease specific guidelines, professional best practices, and associated behavioral research informed the evidence that was translated into the intervention design. Systematic application of these domains resulted in an intervention plan that was used to guide decisions about product features and content.

Behavioral Domains, Essential Behaviors, Supporting Actions and Determinants

Definition

Behavioral domains are the categories of self-management behaviors, and encompass essential behaviors, their associated supporting actions, and determinants. An essential behavior is a behavior "...that should be emphasized through program interventions because of its impact on public health, its measurability, and its feasibility to be performed by patients, caretakers and/or health workers" [17]. Ideally, research has demonstrated that the behavior is associated with improvements in clinical outcomes or longer-term health impact or quality of life. Supporting actions, or sub-behaviors, are the combined small, do-able actions that comprise the essential behaviors [31]. Determinants identified are predictors of behavior change or of present behavior [32].

Derivation

Based on the program objectives for key stakeholders, standards and guidelines were used to inform the identification of behavioral domains supporting selected clinical outcomes (eg, American Association of Diabetes Educators 7 behavioral domains; AADE7) [21]. When available, published research demonstrating correlations of behaviors with clinical outcomes [19] was utilized to identify essential behaviors. Interdisciplinary teams of clinical, behavioral, and user interface/user experience (UI/UX) SMEs identified a broad set of supporting actions for each essential behavior and noted specific actions that could be supported through mHealth interventions. Finally, theory, published literature, formative research, and marketing research provided insights on behavioral determinants.

Multidimensional Profile

Definition

The multidimensional profile is a segmentation approach. It drives the individualization of a user's experience through the customized delivery of interventions via features and tailored message content. Customized delivery can be achieved via preferences set by users or automated, dynamic, and adaptive delivery of interventions based on analysis of longitudinal patterns of data [8]. Content tailoring includes targeted messages based on demographic characteristics, personalization (eg, incorporating first name into messages), or tailoring content in response to assessments, captured data, or the context in which data was captured (eg, bedtime blood glucose [BG] reading) [33].

Derivation

Once goals and essential behaviors were determined, formative market research and the published literature informed the various dimensions of a user profile that were relevant to the essential behaviors. Dimensions included but were not limited to clinical, behavioral, psychosocial, contextual, and personal factors.

Evidence-Based Clinical/Behavioral Interventions

Definition

Evidence-based interventions are those that have been identified as effective for achieving outcomes, and are best practices,

which have been peer-reviewed and evaluated for effectiveness in improving health outcomes [34].

Derivation

These interventions were based on program goals for stakeholders, essential behaviors, and user profiles, and whenever available, the most rigorous quality of evidence was utilized [34]. Interventions were first identified through an analysis of clinical guidelines, standards of care, evidence-based public health programs, medicine, and health care, as well as meta-analyses and systematic reviews of intervention research. However, given the emerging nature of mobile technology, other types of evidence were also considered, including state-of-the-art practices, and best and emerging practices for innovations [34]. Interventions were categorized based on their strategic intent (eg, monitoring support or education support) for incorporation into the framework [35].

App Feature/Content

Following the "waterfall" process, intervention plans were then translated into product features and content identified to deliver optimal outcomes to meet product/program objectives.

Definition

Product features, functionality, and content are designed to deliver program interventions in the most effective means possible, by leveraging state-of-the-art technology with the goal of a highly engaging user experience. Content includes self-management educational curriculum, defined as a coordinated set of educational experiences with specific learning outcomes and employing teaching strategies that are dynamic and reflect current evidence and best practice guidelines [20]. Message content is derived from the educational curriculum, may target the intended behavior, and appeal to the user. Messages may address key benefits and potential obstacles as well [36].

Derivation

Features, functionality, and content incorporated into the app were informed by the aforementioned domains of the framework and developed using an iterative design process. Features and functionality evolved with technology advances, but were always guided by the objectives and intent of the defined set of interventions. New innovations were also considered if it appeared that they could facilitate intervention objectives. For example, a GPS-facilitated feature to identify nearby restaurants and their menus to support meal planning and healthy eating was added to the diabetes app. Table 1 provides examples of the intervention plan that informed the design of product features for the diabetes app.

Table 1. Diabetes mHealth interventions mapped to features.

Intervention type	Intervention description	Product features/content
Educational and skills-building support	Educational and skills-building curriculum content that can be delivered universally to all issues or customized based on individual user data.	Universal education videos, tips
Monitoring support	Guidance for structured blood glucose monitoring with regards to activity types, timing, and frequency of data collection and visual displays of data collected	Logbook (journal blood glucose values, carbs, physical activity); structured blood glucose checking feature
Coaching support	Tailored real-time feedback and trending messages based on customized care-plan prioritized behaviors.	Real-time feedback; longitudinal feedback; customized delivery of video content
Behavioral adherence support	Behavioral adherence tool-set pushed to support customized-care plan. Adherence tools may be accompanied by coaching to address associated self-identified barriers to and motivators for action.	Medication adherence tools (medication list, medication schedule, medication reminders); carb estimation tool; restaurant locator
Patient-provider communication support	Reports sent to health care team with tailored content based on system analysis (eg, patient data inputs and specific provider recommendations) to facilitate patient-provider discussions.	Tailored health care provider report
Patient engagement	Product features, functionality, and content developed specifically to encourage product use.	Homepage design; time-based "touchpoint" messages

Application and Refinement of the Framework

The framework was applied to the development of integrated clinical/behavioral interventions for use in the development of mobile apps for 7 chronic diseases (diabetes, epilepsy, asthma, chronic obstructive pulmonary disease, lupus, HER2+ breast cancer, and low back pain). The findings from this process served as the basis for a chronic disease mHealth intervention taxonomy for scalable design. Table 2 lists the details of the taxonomy. The attributes of the domains were informed by market research with various groups (health care providers, patients, SMEs, caregivers, large pharma) and evidence-based literature (national clinical guidelines, peer-reviewed articles, best practices) that were determined to be valuable in the management of chronic disease.

Table 2. Chronic disease mHealth app intervention design taxonomy.

Domains	Attributes
Value drivers	Clinical; behavioral; psychosocial (quality of life); health care costs; patient engagement in their healthcare
Outcomes and metrics	Clinical; behavioral; quality of life; intermediate outcomes (eg, knowledge); patient engagement
Key stakeholders and program objectives	Patient; health care team; caregiver; social support community; improve diabetes self-management; improve clinical decision-making; improve patient-provider communication
Essential behaviors	Medication-taking; monitoring; problem solving; eating; reducing risk (complications); being active; healthy coping; patient engagement; patient-provider communication
Multidimensional profile	Clinical; behavioral; psychosocial
Clinical/behavioral interventions	Education and skills-building support; monitoring support; coaching; behavioral adherence support; time management support; problem-solving support; patient-provider communication support; social support
Features/functionality/content	Logbook/journal/tracker; self-management tools; reminders; alerts; calendar/scheduling features; patient-provider discussion guide; patient data summary reports; online social network; interactive tutorials/guidance
	Educational content: standard (guideline-based content); customized
	Message content: prompt time-based; real-time feedback; trending feedback

In total, 5 principal programmatic value drivers and 3 program objectives for key stakeholders emerged. There were variations in patient self-management objectives, ranging from single aspects of self-management to comprehensive self-management programs.

Informed by the American Association of Diabetes Educators behavioral domains (AADE7), a total of 7 health behavioral domains were validated across diseases. However, specific essential behaviors facilitated by the mHealth product within each of the domains were specific to the disease (eg, self-monitoring of blood glucose versus self-monitoring of seizure activity). Two additional patient behavioral domains emerged from the process: (1) a health behavioral domain (engaging in health care visits/communicating with the health care team), and (2) initial and ongoing engagement in mHealth product use, which emerged as critical from a program implementation perspective. An analysis of the multidimensional profile for appropriate segmentation of delivered interventions and content yielded 3 principal profile dimensions to be addressed during the first stages of program implementation.

Overall, 8 core evidence-based intervention types identified in the literature were validated via the iterative product development cycle. Additional interventions were incorporated into strategies based on formative and Voice of the Customer marketing research. For example, for the epilepsy app, participants were specific about avoiding the term “epileptic”; for the HER+2 cancer app, participants only wanted information specific to their type of cancer and not breast cancer in general. Also, any participants linking to a social community wanted the participants of that community to have the same specific disease (eg, type 2 versus type 1 diabetes). Patients indicated the importance of these interventions for ongoing engagement with mHealth products. Interventions could include messages that were based on clinical guidelines and/or simply designed to enhance engagement, depending on condition and program objectives. For example, in the diabetes app, real-time feedback messaging served to provide clinically relevant content for the user, and in the epilepsy app, it was used to engage the user in tracking information.

Using the Finalized Framework for Product Development

The “waterfall” framework was applied to the design of an FDA-cleared diabetes prescription app for type 2 diabetes self-management. Table 3 links the design elements to the framework domains.

Table 3. Application of the framework to the mHealth prescription product (stakeholder input on the Diabetes mHealth App)[a,b].

Framework domain		Design elements
Value drivers		Clinical behavioral engagement
Outcomes and metrics		
	Clinical[c]	*Outcome:* At target or improved *Metrics:* A1c<7% or 8% (risk) or 1% improvement if above; BP<140/90 or decrease 10/5 mm/Hg if above; LDL<100 mg/dL or <70 (cardiac risk) or a decrease of 26% if above; HDL>50 mg/dL for females or an increase of 8% if above; HDL>40 mg/dL for males or an increase of 8% if above; Triglycerides<150 mg/dL or a decrease of 26% if above
	Behavioral[d]	*Outcome:* Improve or maintain medication adherence *Metric:* Medication adherence: ≥80% adherence to metabolic meds (correct meds and as prescribed)
	Patient Engagement[e]	*Outcome:* Initial mHealth application use. Sustained mHealth application use *Metrics:* One interaction with the application each week for 1st 3 months
Program objectives for stakeholders		Effective diabetes self-management; Effective clinical decision-making; Effective patient-provider communication
Essential behavioral domains/supporting actions		AADE7[f]; medication-taking; monitoring (BG[g]); problem-solving (high & low BGs); eating; reducing risk (complications); being active; healthy coping; engagement
Multidimensional profile		
	Clinical	Medication regimen; BG value ranges
	Contextual	BG reading type
	Behavioral	Medication-taking behaviors: consistent, inconsistent, nonadherent
EBG-driven clinical and behavioral interventions (evidence-based)		Educational and skills-building support; self-monitoring of blood glucose support; coaching support; behavioral adherence support (medication-taking, carb counting); patient-provider communication support; problem-solving support (eg, addressing high and low BGs); social support
App features/content		
		Logbook; medication list & schedule; carb estimation tool; SMBG[h] tool; reminders; alerts; patient data summary report for health care team; clinical decision support feature
	Information/educational content	Learning library; ADA[i] standards of care; DSME/S[j] educational topics; content to support AADE7 self-management behaviors; tips
	Message types	*Reminders Time-based "touch point messages":* daily messages for engagement, motivation *Longitudinal feedback messages:* based on multiple BG data points entered into journal to improve skills for SMBG *Real-time feedback:* Based on data entered into journal to improve skills for SMBG

[a] Implementing organization = health care technology company

[b] Program implementation model = Rx only by health care providers for patients

[c] Clinical outcomes for chronic conditions (at the patient level)

[d] Behavioral outcomes related to patient self-management

[e] Initial and sustained mHealth application use

[f] AADE7: American Association of Diabetes Educators behavioral domains

[g] BG: blood glucose

[h] SMBG: self-monitoring of blood glucose

[i] ADA: American Diabetes Association

[j] DSME/S: Diabetes self-management education and support

Discussion

The framework resulted in a systematic, replicable, and scalable mechanism for designing mHealth product features, functionality, and content to improve health outcomes in real-world settings for a range of chronic diseases. Through iterative research, design, and testing processes, the "waterfall" framework was created to provide a mechanism for translating evidence and research across multiple disciplines. This framework can be used to drive the systematic development of apps designed to provide different levels of intervention support,

from tracking to comprehensive care management for various diseases.

In order for mobile technology to achieve its promise of revolutionizing health care, we must determine which aspects of mHealth work, for which diseases, for whom, and to achieve which outcomes. This framework can provide a standard approach to design and evaluate the effectiveness of health care apps and to inform policy, practice, and research.

From a health care policy and regulation perspective, if mobile technology is to be used as a form of therapy, it is critical to have a framework that translates existing guidelines and practices into mHealth products in a transparent, universal, and standardized way. Such a framework creates a mechanism that connects guidelines, health care practices and programs, user interface and experience, and mobile technology capabilities for the design of mHealth products.

The development of the framework also shifted the paradigm from feature-based to programmatically driven mHealth design, providing interdisciplinary product teams with a common understanding of the goals and objectives of product features and functionality. The framework influences decisions related to UI/UX, and how to ensure a user experience that integrates support into a person's daily self-care activities. Finally, the framework has the potential to adapt to evolving health care service delivery systems, informing the design of interventions that drive outcomes which support new initiatives.

The framework offers an avenue for researchers to understand how and where guidelines, standards of care, and evidence can be advantageous for mHealth design. It facilitates iterative design and testing during the concept development phase, and process and impact evaluation, which can be used to inform future design. Future applications should validate the framework in diverse service delivery settings so that it can be refined based on broader utilization.

Mobile health technology creates a shift in the paradigm of chronic disease management. It offers new possibilities to engage patients in self-management of their chronic diseases in ways that did not exist in the past. To maximize the potential of mHealth requires the integration of research and expertise from multiple disciplines including clinical, behavioral, data analytics, and technology to achieve patient engagement and health outcomes. This paradigm shift also triggers a need for new approaches to designing clinical and behavioral support for chronic disease management that can be implemented through existing health care services and programs.

The Chronic Disease mHealth App Intervention Design Framework domains and the corresponding design elements developed through this process may provide a foundation for the digital health industry to systematically expand mobile health interventions and validate their effectiveness across multiple implementation settings and chronic diseases. Further enhancement and validation of the framework is needed to recognize these benefits.

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Authors' Contributions

CCW contributed to the conceptualization of the overall platform and frameworks associated with clinical aspects of chronic disease management. RCAK contributed to the conceptualization of the overall behavioral framework and associated platform components. CCW and RCAK contributed to the programmatic delivery of clinical/behavioral interventions through mobile technology, and the writing of those aspects in the paper. MP contributed to the conceptualization of the AADE7 self-management behavioral outcomes framework and associated translation of the informatics components of the taxonomy and associated platform components, and contributed to the writing and editing of those components.

Conflicts of Interest

CCW and MP are employed by, have stock options in, and were supported during time spent writing this article by WellDoc. RCAK was employed by WellDoc when the work described in the publication was being implemented, and has stock options in WellDoc.

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Abbreviations

AADE7: American Association of Diabetes Educators seven behavioral domains
BG: blood glucose
FDA: Federal Drug Administration
mHealth: mobile health
SME: subject matter experts
UI/UX: user interface/user experience

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Original Paper

Improving Patient Experience and Primary Care Quality for Patients With Complex Chronic Disease Using the Electronic Patient-Reported Outcomes Tool: Adopting Qualitative Methods Into a User-Centered Design Approach

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Abstract

Background: Many mHealth technologies do not meet the needs of patients with complex chronic disease and disabilities (CCDDs) who are among the highest users of health systems worldwide. Furthermore, many of the development methodologies used in the creation of mHealth and eHealth technologies lack the ability to embrace users with CCDD in the specification process. This paper describes how we adopted and modified development techniques to create the electronic Patient-Reported Outcomes (ePRO) tool, a patient-centered mHealth solution to help improve primary health care for patients experiencing CCDD.

Objective: This paper describes the design and development approach, specifically the process of incorporating qualitative research methods into user-centered design approaches to create the ePRO tool. Key lessons learned are offered as a guide for other eHealth and mHealth research and technology developers working with complex patient populations and their primary health care providers.

Methods: Guided by user-centered design principles, interpretive descriptive qualitative research methods were adopted to capture user experiences through interviews and working groups. Consistent with interpretive descriptive methods, an iterative analysis technique was used to generate findings, which were then organized in relation to the tool design and function to help systematically inform modifications to the tool. User feedback captured and analyzed through this method was used to challenge the design and inform the iterative development of the tool.

Results: Interviews with primary health care providers (n=7) and content experts (n=6), and four focus groups with patients and carers (n=14) along with a PICK analysis—Possible, Implementable, (to be) Challenged, (to be) Killed—guided development of the first prototype. The initial prototype was presented in three design working groups with patients/carers (n=5), providers (n=6), and experts (n=5). Working group findings were broken down into categories of what works and what does not work to inform modifications to the prototype. This latter phase led to a major shift in the purpose and design of the prototype, validating the importance of using iterative codesign processes.

Conclusions: Interpretive descriptive methods allow for an understanding of user experiences of patients with CCDD, their carers, and primary care providers. Qualitative methods help to capture and interpret user needs, and identify contextual barriers and enablers to tool adoption, informing a redesign to better suit the needs of this diverse user group. This study illustrates the value of adopting interpretive descriptive methods into user-centered mHealth tool design and can also serve to inform the design of other eHealth technologies. Our approach is particularly useful in requirements determination when developing for a complex user group and their health care providers.

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KEYWORDS

eHealth development; mobile apps; multi-morbidity; complex patients; primary care

Introduction

Overview

Software developers have historically relied on standard processes for requirements determination and functional specification [1,2]. Commonly used development models range from the conventional waterfall approach (ie, sequential design process) [3] through to more contemporary methodologies such as agile development (ie, frequent testing of iterative designs) [4]. The evolution in methods for functional specification have largely focused on finding ways to mitigate the challenge created by user or environment requirements that evolve during the development lifecycle. Iterative development approaches such as prototyping emerged to address difficulties experienced in identifying user requirements.

Almost all software requirement elucidation techniques assume the ability to engage—at specific stages or in all stages in the software specification process—with cognitively and physically able user populations. Less well understood is how to successfully elicit user specifications from medically fragile populations, such as those with complex chronic diseases and disabilities (CCDDs) [5-7]. These are individuals with two or more chronic conditions (ie, multi-morbidity) and who often face social, environmental, and contextual issues that impact on their health care needs and ability to manage [8]. These patients are among the heaviest users of the health system [7], and thus increasingly garner the attention of insurers—public or private—who are searching for cost-effective solutions [9]. At the same time, patients are seeking to minimize the burden of their illness as they navigate their way through a highly complex health care system.

In today's technology-enabled world, there has been a plethora of patient-centered apps, online medical resources, wearable fitness technologies, and similar tools to promote adherence to treatment plans [10-13]. What is strikingly absent are information-enabled solutions designed specifically for high users of the health system [14], like patients with CCDD. The majority of apps are focused on single-disease management, thereby not aligned with the needs of complex patients [15]. Furthermore, development methodologies used in the creation of eHealth solutions lack the ability to embrace users with CCDD in the specification process.

We posit that eHealth solutions that target CCDD patients are needed, and that the success of these solutions will in part be determined by incorporating persons with CCDD in the development process. We adapted a conventional software development process to make CCDD patient users central to the specification and testing process. Using a multi-phased user-centered approach, we sought to build, deploy, and test a patient-centered app to improve quality of care and patient experience for patients with CCDD in primary health care settings. This paper describes the development phase in the creation of the electronic Patient-Reported Outcomes (ePRO) tool, with particular attention to the use of qualitative methods incorporated into the software specification process to facilitate user feedback from CCDD patients—a user population for which traditional design approaches may not be well tailored. Tool development phases were supported by the technology partner, QoC Health Inc.

Background and Significance: Adopting Qualitative Methods in a User-Centered Design Approach

Until the mid-1990s, most software development methodologies were anchored in a linear process based on the premise that the more detailed the specification, the greater the prospect of realizing a well-functioning solution. The System Development Life Cycle [16], IBM's Joint Application Design (JAD) [17,18], and later the Rational Unified Process [19] were typical of the era. As the opportunities to "computerize" increasingly complex systems grew, development methodologies shifted toward increasingly iterative development processes, with agile development being an excellent example of a contemporary approach [4]. Central to these approaches was the concept of user-centered design [20].

User-centered technology development is "characterized by a focus on the user, and on incorporating the user's perspective in all stages of the design process" [21, p 1]. This approach is often iterative, involving multidisciplinary design teams, and emphasizes the need to incorporate user feedback as part of the design, testing, and implementation process [21]. Previous studies have noted that adopting user-centered design approaches can improve usability and implementation [22] and can also improve user acceptance and satisfaction with new systems overall [23]. Iivari and Iivari [24] suggest that there are four dimensions of user-centeredness: user focus, work-centeredness, user involvement or participation, and system personalization. One or more of these dimensions may be the focus on the user-centered method employed, and it is suggested that developers and researchers strive to include all four in the design approach. Also important is attention to the provision of appropriate processes and supports to encourage meaningful

engagement and empowerment by users involved in the design process [25].

Design evaluation approaches suggest the use of rigorous research methods and evaluations in order to support capturing and incorporating user input [26]. We looked to combine rigorous qualitative research evaluation with an iterative design approach to obtain user feedback to develop a mobile solution that will meet the needs of patients and their primary care providers. Qualitative methods that capture individual experiences and perceptions [27] provide a useful toolkit to a user-centered design evaluation approach. Interpretive description, a form of qualitative inquiry, draws on data collected through in-depth interviews and focus groups to capture human experience [28]. Understanding the care experiences and needs of patients with CCDD requires attention to the context in which their physical and mental health and social needs are intertwined [8]. Qualitative inquiry captured in an open setting closer to participants' lived experience allows us to capture the complexity and breadth of experiences of patients with CCDD, which may not be possible through typical user-centered design approaches, such as one-on-one, lab-based walk-throughs [29], which capture information—sometimes both qualitative and quantitative—in a closed setting. Iterative changes to the tool can be informed by rich user experience data—ensuring user focus—supporting user involvement in the design and development of a truly person (user)-centered tool.

This paper describes the design and development approach to create the ePRO tool, specifically the process of incorporating qualitative research methods into user-centered design approaches. Key lessons learned are offered as a guide for other eHealth and mHealth research and technology developers working with complex patient populations and their primary care providers (PCPs).

The Electronic Patient-Reported Outcomes Tool

In this section, we offer a brief description of the ePRO tool that was developed through the process described in this paper. The ePRO tool includes a portal system for providers and patients to set up and monitor goals, as well as a mobile device for patients to track their goals. There is also a Hospital CheckOut feature that allows patients to report when they have visited and been discharged from a hospital—primary care providers see this information when they access the portal. To set up goals, patients and providers collaboratively work together on the portal system during an in-person visit. After initial set-up, both patients and providers can view patient progress on the portal any time in between visits. Patients also have the option of inputting their monitoring data on the portal if they do not wish to use their mobile device. A full description of the new tool will be published in our forthcoming paper outlining our usability pilot (see Multimedia Appendix 1).

Methods

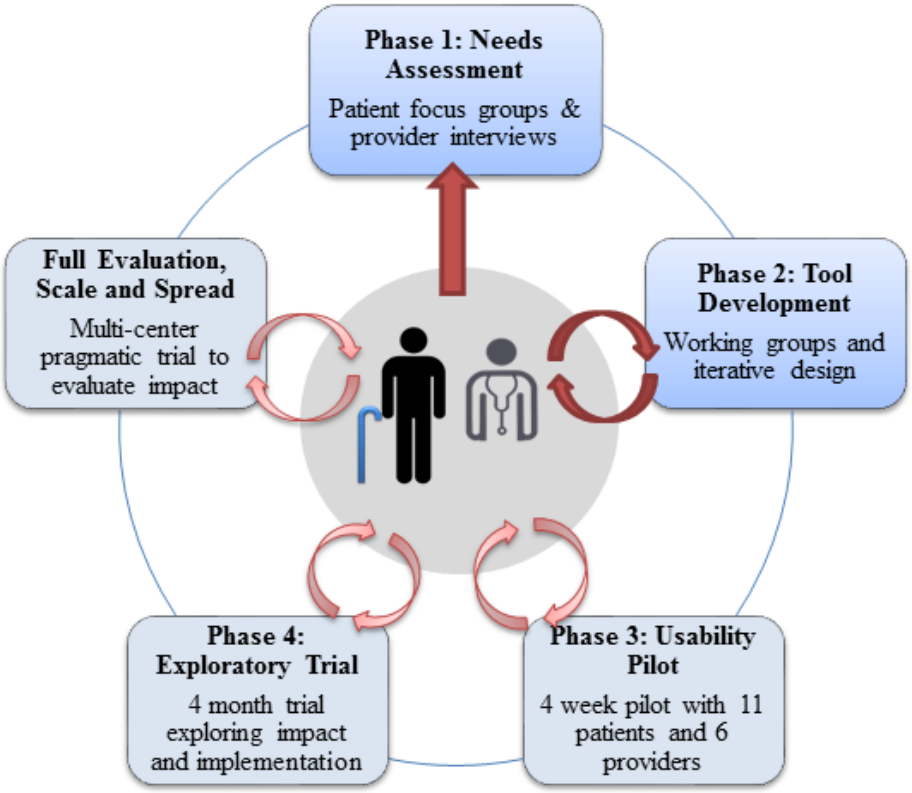
Overview

Guided by user-centered design principles [21] and other studies involving the development of mHealth technologies [30-33], we adopted a multi-phased research approach to support the iterative design and development of the ePRO tool. Full ethics approval was obtained from the Joint Bridgepoint Hospital-West Park Healthcare Centre-Toronto Central Community Care Access Centre-Toronto Grace Health Centre Research Ethics Board. At the outset, we broadly intended to develop an mHealth solution to support community-dwelling patients with CCDD and their PCPs. Figure 1 provides a visual depiction of our design and development method.

As depicted in Figure 1, patients, carers, and PCPs were involved at each step in the design process. This paper describes Phases 1 and 2 with specific emphasis on provider input, as shown in Figure 1. Methods and findings from the patient-focused aspects of Phase 1 have been published elsewhere [34,35].

The tool was developed within a primary health care practice, which included an interprofessional team of PCPs composed of physicians, nurse practitioners, registered nurses, social workers, and dietitians. Feedback on the tool was captured from PCPs who varied in their interest and willingness to engage in new technologies. Two PCPs expressed low interest and willingness in the working group, while the rest were more keen to try new technologies as part of care delivery.

Figure 1. Development approach.



Phase 1: User Needs Assessment—Provider and Expert Input and Response to Patient-Identified Needs

Phase 1 consisted of focus groups with patients and carers plus interviews with content experts (CEs) and PCPs from the practice. This phase of development was mainly concerned with ensuring a user focus and capturing work-centeredness dimensions of user-centered design in that we sought to understand users' needs and the tasks needed to address those needs. Focus group findings in Phase 1, along with a literature review [34], provided the initial building blocks for tool development. A total of 14 patients and carers participated in four focus groups in the fall of 2013. Of the 14 participants, 10 (71%) were patients, 2 (14%) were caregivers only, and 2 (14%) were both patients and caregivers. The participants' average age was 64 years (range 42-90), and 9 out of 14 (64%) participants were female. Patients participating in the focus groups reported multiple chronic illnesses, including diabetes, chronic pain, osteoarthritis, osteoporosis, anemia, cardiac conditions, glaucoma, and mental illness [35]. Initial findings from the focus groups and available resources suggested that the tool should support better communication between patients and PCPs around three key areas:

1. Information about symptoms and functional status (ie, pain, mobility, depression/anxiety, activities of daily living [eg, bathing, toileting], and social well-being).

2. Medication management support (ie, reminders, renewals, and reporting side effects).

3. Educational materials and/or trusted websites to support self-management.

Next, purposive sampling [36,37] was used to identify PCPs and CEs who could provide the feedback required to refine the tool. Semistructured interviews were conducted with the PCPs, as well as CEs, in at least one of the fields of development or utilization of eHealth and/or research or service delivery experience with CCDD patients. CEs were identified through their academic, clinical, and/or research networks.

PCPs were selected from the primary care practice where the tool would be piloted and tested. These PCPs had been engaged in the project from early stages and had attended several meetings to receive updates on the project. A patient advocate was also interviewed, as this individual is experiencing CCDD who has engaged with other eHealth technologies as part of their care, and has previously served as a patient representative in other research projects.

PCPs and CEs were given a summary of patient focus group findings and asked their perspectives on the following: (1) the value of ongoing monitoring of symptoms and functional status as part of usual care, (2) what types of information should be shared about those symptoms (ie, indicators, scales, and contextual information) and how it could best be shared, (3) the role of patients accessing appropriate educational materials, (4) how different communication methods would fit into provider workflows, and (5) other aspects of primary health care delivery that may be important to capture for managing patients with CCDD.

Phase 2: Tool Development

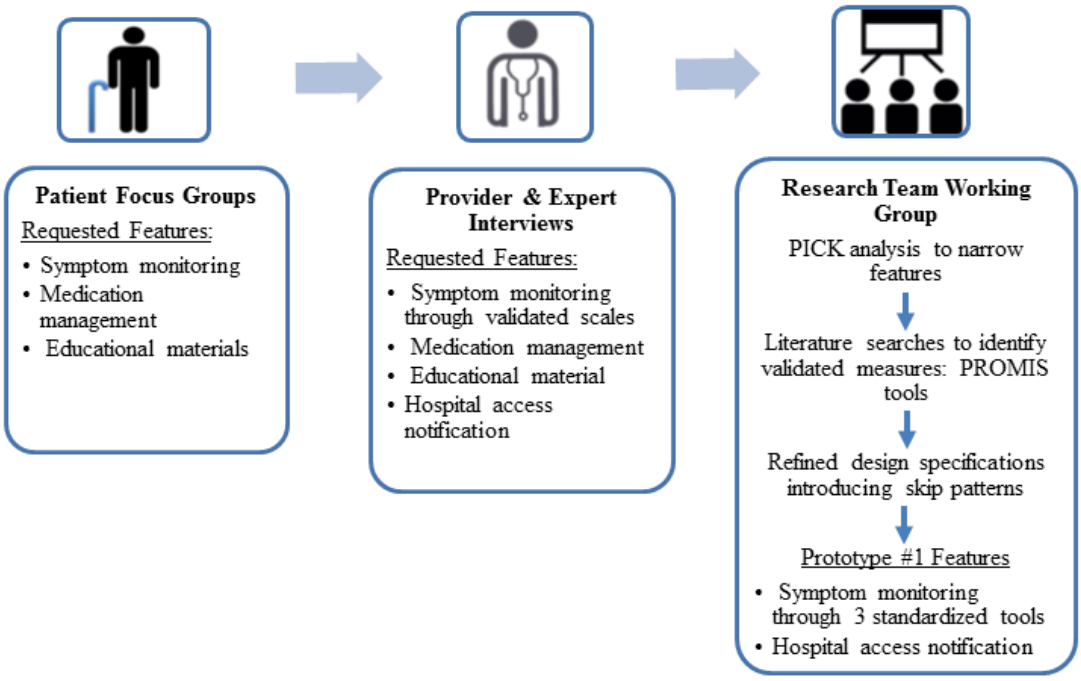
Based on the Phase 1 findings, we identified four key features for the tool: symptom monitoring; medication management; educational resources; and hospital visit notification, as hospital access notification was identified in the provider interviews as

an ongoing problem. Figure 2 shows the prototype development process; in the figure, the first two icons are open source from Iconfinder [38] and the last icon is open source made by Flaticon [39]. Prototype development and refinement occurred over a series of teleconferences and meetings between the research partners and the technology partner.

Patients, PCPs, and CEs who participated in the earlier stages of the study were invited to provide feedback on a working prototype during separate 2-hour working group sessions. At each working group session, participants were asked (1) whether the tool captured issues of importance to patients with CCDD, (2) whether the tool was easy to use and understand in terms of question wording and interface, and (3) whether there were other ways that we could gather similar information. The working groups aimed to support user involvement in the design process, while allowing us to pay attention to unique use cases and identify how we could build in system personalization.

The working groups consisted of modified cognitive walk-throughs [29]—participants "walk through" the sequence of tasks—in order to test the usability of the system. In contrast to one-on-one cognitive interviews, working groups provide an opportunity for individuals to engage in open dialogue and reflect on their diverse personal and professional experiences allowing us to effectively capture the breadth of multiple user experiences. Each session was audiotaped and recordings were used to verify and substantiate researcher notes taken during and after the groups.

Figure 2. Prototype development process. The first two icons are open source from Iconfinder [38] and the last icon is open source made by Flaticon [39]. PICK: Possible, Implementable, (to be) Challenged, (to be) Killed.



Data Analysis

Data analysis was conducted by reviewing notes and transcripts from interviews and working groups. An iterative analysis technique was used in which data were reviewed by two researchers, first independently and then together at multiple points aligned with the stage of tool development. Findings were organized in relation to the tool design and function to help systematically inform modifications to the tool. We used an interpretive descriptive approach in which findings are compared to a starting point [28]—the ePRO tool and prototype. The tables were written up into a summary report for Phase 1 (PCP and CE interviews) to inform the development process described above, and another for Phase 2 (prototype development process) to inform additional tool changes to be made prior to Phase 3 (usability pilot to be described in a future manuscript).

Results

Participants

Table 1 offers a description of the participants in Phases 1 and 2. Patients and carers who participated in the Phase 2 working groups had also attended the Phase 1 focus groups. Although more patients and caregivers from Phase 1 were invited to participate in Phase 2, many were unable to attend due to health issues.

Phase 1: Primary Care Provider and Content Expert Interviews

PCP and CE interviews ran for approximately one hour. PCPs and CEs identified that symptom monitoring, medication management, and educational resources were all important aspects of care delivery for patients with CCDD. All supported the idea of ongoing monitoring of pain symptoms and mobility in relation to activities of daily living, as well as anxiety and depression symptoms. PCPs and CEs identified a variety of

symptom-related variables and scales with little consensus except for the need for validated measures. PCPs identified the need to know when their patients were admitted to a hospital, as they frequently did not know when this had occurred, resulting in little or no follow-up and poorly coordinated care.

Phase 2: Prototype Development Process

Figure 2 depicts how the user needs assessment was used to inform the iterative prototype development process. As can be noted in Figure 2, two features were removed through the design process by engaging in a PICK analysis—PICK stands for Possible, Implementable, (to be) Challenged, (to be) Killed. A PICK analysis is a lean technology development process in which features are assessed in terms of the time and resource investment required, and the anticipated value added [40]. The assessment allowed us to determine whether a feature was (1) Possible (ie, easy to accomplish but with a low value add), (2) Implementable (ie, easy to accomplish with a high value add), (3) to be Challenged (ie, was difficult to accomplish but had a high value add), or (4) to be Killed (ie, difficult to accomplish with a little value add). Our focus was on identifying features that fell into the *Implementable* category. The medication management and educational features were challenged, as they were difficult to accomplish given our resources and time frames. In addition, our technology partner was developing similar features for another project, thus the features could be incorporated into the ePRO tool in a future iteration.

The prototype included two features: symptom monitoring and hospital access notification. Symptom monitoring focused on symptoms identified as most important in the patient focus groups—pain, mobility, anxiety/depression, and social well-being [35]. A literature review revealed three Patient-Reported Outcome Measurement Information System (PROMIS) tools—the Global Health Scale, the Pain Interference Scale, and the Improved Health Assessment Questionnaire—as appropriate to needs as they captured symptoms of interest and were validated in chronic disease populations [41-43]. To minimize respondent burden, skip patterns were created so patients could only answer scales that were relevant to their current symptoms. A free-text comment box was included at the end of the symptom reporting to allow patients to provide contextual information to meet the needs of patients who wanted their providers to understand them as “whole persons” [35].

The hospital visit notification feature allowed patients, and/or their carers, to notify the PCPs when the patient had been to an emergency department or admitted to hospital, including the name of the hospital, date of admission, and reason for admission. Once notified, PCPs could request discharge reports from hospitals, which are not typically received in a timely manner.

Working Group Findings: A New Direction

The three working groups provided rich and valuable feedback on the prototype. Findings were separated into categories of *what works* and *what does not work*.

What Works

Functionality: Ongoing Monitoring and Tracking

All three groups felt the remote monitoring function was valuable. Patients appreciated being able to see changes in their symptoms over time. PCPs felt that having information about their patient’s symptoms over time could enable them to see what specific issues their patients had been facing and then target discussions at the point of care to those issues.

Content: Including Contextual Information

All three working groups saw value in capturing contextual information through the use of open-ended questions in order to provide a more well-rounded understanding of patients' symptoms and capacity to self-manage.

What Does Not Work

Issue 1: The Length of the Tool

All three groups felt that the monitoring questions were overly burdensome. For patients, this meant potentially taking 20-25 minutes once a week to answer questions, and for PCPs it entailed sifting through a large amount of monitoring data.

Issue 2: Fitting in With Provider Workflows

While PCPs saw the potential for symptom monitoring between visits, they were uncertain how they would fit monitoring into their daily schedules. PCPs also had concerns about liability issues, if they were to be responsible for monitoring a large number of patients.

Issue 3: Unclear Answer Keys, Scales, and User Flow

There were concerns from all three groups regarding confusing visual analog and Likert scales. Patients expected a rating of 10 to be good; however, the scales were based on tools designed for providers, who tend to see high values (ie, spikes) as a negative health outcome. Additionally, some of the scales were flipped. A high number would be a good outcome for one question but a bad outcome for another. It was felt that standardization in line with the preference of the patients would improve usability of the tool.

Table 1. Study participants.

Participants	Details	Type of group in each phase	
		Phase 1	Phase 2
Patients	9 female Average age=64 years (range 42-79) 2 patients were also caregivers All identified have two or more chronic conditions they find difficult to manage	Focus group (n=12)	Patient/caregiver working group (n=4) 3 females, 1 male Of these, 1 female patient was also a caregiver
Caregivers	All female	Focus group (n=2)	Patient/caregiver working group (n=1)
PCPs [a] (n=7)	General practitioners (n=2)	Interview	Provider working group
	Nurse practitioner	Interview	Provider working group
	Registered nurse	Interview	Provider working group
	Dietitian and diabetes educator	Interview	Provider working group
	Administrative staff member	Interview	Provider working group
	Executive director	Interview	N/A[b]
CEs [c] (n=6)	Complex pain	Interview	Expert working group[d]
	eHealth	Interview	N/A
	eHealth for chronic conditions	Interview	Expert working group
	Rehabilitation in complex populations and PROs[e]	Interview	Expert working group
	Complex stroke	N/A	Expert working group
	Complex patient	Interview	Expert working group

[a]PCP: primary care provider.

[b]N/A: not applicable.

[c]CE: content expert.

[d]Research team participants in expert working group (n=4).

[e]PRO: patient-reported outcome.

Issue 4: Content Issues

The wording of some questions, including length, reading level, double-barreled questions, and negative labeling connotations for some questions—particularly around mental health—were seen as problematic by patients. Providers also wanted to see questions on patient confidence and self-efficacy included, which were subsequently added to the tool.

Shifting From Monitoring to Supporting Self-Management and Patient-Centered Delivery

The issues identified by all participants suggested the need to rethink the utility and purpose of the tool moving forward. Provider workflow concerns, the length of the tool, and content issues suggested very low usability of the prototype. More importantly, working group findings revealed that much of the symptom data of importance to patients were related to the types of care planning and goal setting that the PCPs would engage in with patients. This realization prompted a major shift in the purpose of the tool away from an application that only captures patient-reported outcome measures to a tool that actively uses those measures to improve the design and delivery of goal-oriented primary health care while supporting self-management for patients with CCDD.

Goal-oriented models of care can support improved patient-centered care delivery [44], identified as a crucial need via the patient focus groups. Furthermore, a goal-oriented approach to care aligns with the patients' desires to improve their health status with respect to symptom management. Goal setting has been identified as an important process to improve care for complex and chronically ill patients [44,45]. Despite this, goal-oriented care has proven to be a challenge in primary care settings [46] and goals are often not agreed upon between providers and their complex patients [47].

Discussion

Implications for Development

Overview

An interpretive descriptive approach allowed us to capture diverse and unique user needs of a complex and often overlooked patient population. Our data collection and analysis strategy informed a major shift in system requirements which may have been missed if we had relied solely on more traditional requirement elucidation techniques. The discussion will outline three key lessons learned with regard to developing mHealth tools for patients with CCDD in primary health care settings,

and examine how the use of interpretive descriptive methods helped address and respond to these issues. These lessons could extend to development of broader eHealth technologies as well.

Lesson 1: Developing Tools for Patients With Complex Chronic Diseases and Disabilities Requires Balancing Multiple and Diverse Needs

A core challenge in adapting functional specifications methodology for use by people with CCDD was the difficulty associated with balancing the needs of multiple diverse end users. Although the initial intention was to create a patient-centered tool, there had to be acknowledgement of the needs of PCPs who would be engaging with the tool. While the original prototype was meeting patient needs for symptom monitoring, it did not align with provider workflows. Qualitative methods enabled us to capture why individuals responded as they did, including contextual factors that played a role in that response, and allowed for probing at ways to reconcile diverse needs. Codesigning interventions with CCDD patients and providers has already been noted to be key to success [48,49]—an approach that should extend to developing other types of mHealth and eHealth interventions as well.

Other studies have noted the need to engage with users to further refine the purpose and goals of new information technology (IT) systems [30,32]. For example, in designing the Coplintho project, De Rouck and colleagues underwent extensive early development work to identify the appropriate user groups and user needs, as well as to create a final purpose or *use case* for their tool [50]. As is the case in developing the ePRO tool, the Coplintho project drew heavily on qualitative methodologies and the designers concluded that their methods helped to refine project goals and tool functionality.

Lesson 2: The Purpose and Intention of the Tool Should Remain Flexible Through the Development Process

The purpose of the tool had to be flexible in order to meet user needs. Part of the challenge was that there was uncertainty regarding the specific needs of the CCDD patient population, thereby creating a number of design and development challenges, as well as research challenges, since many early discussions around the tool were exploratory in nature. Flexibility allowed for responsiveness to user needs and contexts but resulted in challenges in the research and development process that required resources to assess and continually explore different system capabilities, functionalities, and content.

Lesson 3: Careful Attention Needs to be Paid to Provider and Organizational Barriers to mHealth Tool Adoption

Close attention also needed to be paid to organizational and system realities in which our tool would be adopted. The organizational culture around adopting new technologies required resources in terms of IT support for PCPs. Further, provider time and workflows factored into the provider response and willingness to engage with the tool during development. Through interviews and working groups, these important contextual factors were identified and could be addressed in redeveloping the prototype.

Provider and organizational-level barriers to adopting mHealth technology are well documented in the literature. In a systematic review of factors affecting mHealth adoption, provider-level issues such as perceived usefulness and impact on patient care, and organizational-level factors such as impact on provider workflows and interprofessional communication, were found to be important [51]. It has been recommended that development stages should involve clinical perspectives in order to mitigate barriers and ensure appropriate functionality [14]. If only patient needs were considered, the ePRO tool would serve solely as a monitoring tool, which would have been unsustainable in the long term.

Limitations

Blending qualitative specification elucidation into traditional prototyping methods is time and resource intensive and, as such, we were only able to go through a single round with a high-fidelity prototype which was “an incomplete but essentially executable version of the final product” [52, p 666] . Future studies involving multiple and diverse end users should build in additional time and funding, and potentially use lower-fidelity prototypes like wireframes to enable multiple iterations and cycles of design. Another limitation was the separation of patients and PCPs at the working group stage in order to minimize perceived power differentials that might result in patients not fully expressing their views. This concern may make codesign with both parties in the room difficult [49]. We also had a relatively low number of patients in our working group due to medical frailty. When working with a complex patient population, strategies are required to address the high likelihood of dropout, for instance, recruiting a much larger number of patients for the focus group then would be typically done. Another consideration working with this population is there may be concerns with capturing typical socioeconomic data, such as education, occupation, and household income, which could deter patients from participation. As such, we opted to forgo capturing this information at this stage of development, so as to reduce barriers to participation.

As we move forward with this work, we must also be aware that many individuals within the complex patient population, particularly older adults and those with lower household incomes, may not have their own mobile phones. To address this issue, we will make mobile phones available to participants of our pilots and trials, and make it possible for caregivers to respond on the patient;#8217;s behalf. As noted earlier, patients can also input data via a portal system, which they can access from a home computer or open access computers at libraries.

Conclusions

The use of interpretive descriptive methods can be of particular use in requirements determination and functional specification in software development for diverse populations, such as patients with CCDD. Qualitative methods can do the following: help to ensure all end users' needs are captured and understood within their unique contexts, allow for a flexible purpose of tools in the early stages of design, and help to capture contextual enablers and barriers to the tool’s uptake. The key lessons that have emerged from the process of developing the ePRO tool can be adopted by other researchers or developers of mHealth

and eHealth technologies. Using interpretive descriptive qualitative methods as part of a user-centered design was pivotal to capturing, analyzing, and implementing user feedback. To help generate a better understanding between patients and providers of each other's experiences, future design stages will seek to bring patients and providers together to align with established codesign methods [52].

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Conflicts of Interest

None declared.

Multimedia Appendix 1

Electronic Patient-Reported Outcomes (ePRO) tool screenshots.

[PPTX File, 5MB - resprot_v5i1e28_app1.pptx]

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Abbreviations

CCDD: complex chronic disease and disability
CE: content expert
ePRO: electronic Patient-Reported Outcomes
IT: information technology
JAD: Joint Application Design
N/A: not applicable
PCP: primary care provider
PICK: Possible, Implementable, (to be) Challenged, (to be) Killed
PRO: patient-reported outcome
PROMIS: Patient-Reported Outcome Measurement Information System

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Original Paper

Development and Initial Evaluation of the Web-Based Self-Management Program "Partner in Balance" for Family Caregivers of People With Early Stage Dementia: An Exploratory Mixed-Methods Study

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Abstract

Background: People with dementia increasingly depend on informal caregivers. Internet-based self-management interventions hold considerable promise for meeting the educational and support needs of early stage dementia caregivers (EDCs) at a reduced cost.

Objective: This study aimed to (1) develop an online self-management program for EDC to increase self-efficacy and goal attainment, and (2) evaluate the program's feasibility and report preliminary data on effectiveness.

Methods: Based on the Medical Research Council (MRC) framework for the development and evaluation of complex interventions, a stepwise approach was adopted to explore potential user needs and develop and validate the content by means of (1) focus group discussions with dementia caregivers (N=28), (2) interviews with dementia care professionals (N=11), and (3) individual think-aloud usability tests with EDC (N=2) and experts (N=2). A pilot evaluation was conducted with EDC (N=17) to test the feasibility and establish preliminary effects. Self-report measures of feasibility were completed after the completion of intervention. Self-efficacy and goal attainment were evaluated before and after the intervention.

Results: The different steps provided useful information about the needs of potential users regarding the content and delivery of the program. This resulted in the newly developed "Partner in Balance" program. At the start, system failures resulted in a high noncompleter rate (7/17, 41%), but at the end, an acceptable feasibility score of 209 (range 54-234) was found. The convenience of completing the program at home, the tailored content, and the guidance (face-to-face and online) were appraised positively. Preliminary effects on caregiver self-efficacy (P<.05) and goal attainment (T>50) were promising.

Conclusions: Adaptations were made to the program to limit the amount of system failures and prevent high noncompleter rates. As recommended by the MRC framework, confirming the feasibility and preliminary effectiveness is a valuable step toward examining the effectiveness of this newly developed intervention.

Trial Registration: Dutch Trial Register (NTR): NTR4217; http://www.trialregister.nl/trialreg/admin/rctview.asp?TC=4217 (Archived by WebCite at http://www.webcitation.org/6f6B8lvRP).

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KEYWORDS

carers; dementia; focus groups; Internet; psychosocial support systems

Introduction

Chronic illness and decreased well-being are expected to become global public health challenges [1], with dementia being one of the most common disorders in elderly individuals [2]. With less formal health care available and more people in need of care, the caring role has now shifted to the informal caregivers at home [3]. However, caregivers of people with dementia are at an increased risk of burden, stress, and have a fourfold risk of becoming depressed compared with noncaregivers [4,5]. As such, this transition of friend/family member into the caring role increases the need for effective caregiver interventions to improve their mood and quality of life.

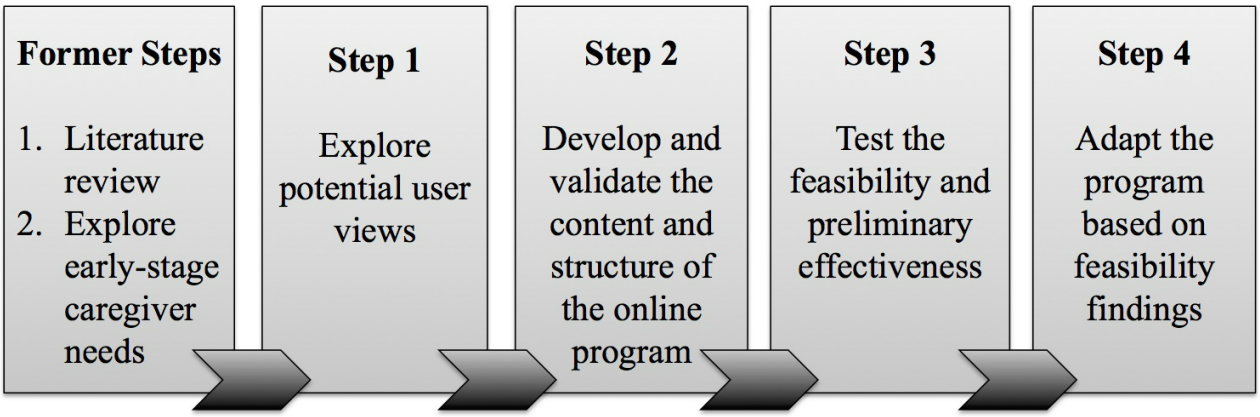
Although recent face-to-face caregiver interventions appeared to be promising [6,7], the increasing gap between care supply and demand calls for alternative and cheaper methods for providing education and support to informal caregivers [3,8]. Internet interventions may help caregivers cope with the challenges of caring for a person with dementia [9]. A recent literature review [10] showed that the currently available Internet interventions for caregivers of people with dementia have promising effects on their confidence and burden, given that they included multiple components and were tailored to the individual participant (caregiver). In addition, Internet-delivered caregiver support may prevent accessibility problems for informal caregivers who are isolated or have difficulties accessing traditional health care services [11,12].

At present, remote support for dementia caregivers is increasing, and new Internet interventions are being developed [13-19]. These programs are, however, mainly focused on dealing with dementia-related problems (eg, neuropsychiatric symptoms) that occur at an advanced stage of the caregiver career; by contrast, early stage interventions can prepare caregivers for their future tasks at a stage where stress and burden are relatively low [20]. Early intervention and support for caregivers have proven to be effective in reducing strain, increasing caregiver confidence, and delaying institutionalization of the person with dementia [21-23]. Moreover, early therapeutic interventions may help caregivers identify their needs based on their individual situation and facilitate the adaptation process [24]. The Stress and Coping paradigm by Lazarus and Folkman [25] and the Social Learning theory by Bandura [26] propose that taking charge of the changes in one's life increases self-efficacy, and can therefore reduce caregiver stress and its impact on general well-being [27]. Following these theories, an early stage support program for caregivers should focus at positively managing life with dementia rather than managing the dementia itself [28]. Self-management programs suit the caring role transition and have previously been used to support informal caregivers of several chronic diseases with promising results [12,29,30].

Iterative Development Process

This study describes the development of an online self-management program for early stage dementia caregivers (EDC) to improve self-efficacy and goal attainment. We closely followed the iterative process of the new Medical Research Council (MRC) Framework for the development of complex interventions [31]. The first 2 steps in the intervention development are described elsewhere [10,24]. The current paper describes the next 4 steps (Figure 1) spread over a 2-year period (2012-2014). These steps are described in the following sections.

Figure 1. Iterative development process informed by MRC framework.



Methods and Results

Step 1: Explore Potential User Views: Focus Groups

Methods

In-depth exploratory focus group interviews were conducted to explore EDC's views on the content and format of an early stage intervention (see Ref [24] for detailed methodology). A context-mapping approach [32] was used: a booklet examining personal Internet and computer use and a collage displaying chosen themes based on significance during the early stages. Available themes were preselected based on existing interventions, early stage dementia care literature [21,33], and expert knowledge. Blank cards were also provided. The most often selected and highest appraised themes were compiled. Focus group interviews were transcribed verbatim and analyzed

independently based on deductive content analysis by 2 of the study authors (LMMB and MEdV). Topics that were mentioned frequently and explicitly served as the basis for categorization. Categories were merged into common themes in a consensus meeting (LMMB and MEdV).

Results

Participant characteristics (N=28) are presented in Table 1. The booklet on computer use was completed by 18 participants. Reasons for noncompletion were (1) overlooking the booklet (N=7) and (2) not understanding its value (N=3). Participants used the computer for multiple purposes, for example, finding information (15/18, 83%), email (14/18, 78%), financial transactions (14/18, 78%), writing (14/18, 78%), viewing photos (9/18, 50%), playing games (8/18, 44%), (video) chatting with family members (3/18, 17%), and shopping (2/18, 11%). However, 3 participants did not use a computer and were not inclined to do so in the near future.

During the focus groups, the majority of the participants considered Internet interventions as efficient due to the high level of accessibility, especially when feeling pressed for time or being bound to one's home. Receiving answers to urgent queries was also considered very positive.

> *The advantages are no travelling time and the possibility to search what I want to know when it is convenient for me.* [P9]

> *I would like to be able to extract the information that is important for me at that particular moment, because we're all so different. When you're in need of an answer, a personal response would be great.* [P13]

Blended care (face-to-face care combined with online modules) was preferred over online care only, due to the personal contact with a professional.

> *People experience emotions, while a computer is just an object. Seeing the person you are talking to is really important. Once you know each other, email or telephone is fine for information exchange.* [P5]

Table 1. Background characteristics of the caregivers (N=28) and the care recipients (N=25).

Characteristics		n (%) or mean
Age, years		63.6
Gender		
	Men	7 (25)
	Women	21 (75)
Relationship to the care recipient		
	Spouse	22 (79)
	Child	2 (7)
	Child-in-law	2 (7)
	Sibling	1 (4)
	Friend	1 (4)
Living together with care recipient		
	Yes	21 (75)
	No	7 (25)
Care recipient diagnosis		
	Mild cognitive impairment	8 (32)
	Alzheimer's disease (AD)	11 (44)
	Vascular dementia	3 (12)
	Parkinson	1 (4)
	Dementia not otherwise specified	2 (8)
Care recipient years of diagnosis		
	0.5	5 (20)
	1-3	9 (36)
	4-6	10 (40)
	7-10	1 (4)

All participants (N=28) completed the collage of themes during the interviews. Participants stressed the importance of an intervention tailored to the stage of the disease and the individual caregiver's situation, with less focus on coping with dementia

and negative stigmatizing information about the future. Learning how to stay healthy by positively managing one's life and learning to accept the changes were considered important, and so is the significance of information provided by other caregivers. A more flexible choice of themes, based on personal needs and areas of interest, was considered desirable. The themes most often mentioned and highest appraised are listed in Table 2.

Table 2. Themes most often selected and highest appraised by family caregivers (N=28).

Themes	Selected n (%)	Appraised most important n (%)
Practical tips	27 (96)	14 (50)
Role and relationship changes	22 (79)	5 (18)
Information about the disease	21 (75)	12 (43)
Balance in activities	19 (68)	2 (7)
Focus on the positive	19 (68)	5 (18)
Communication	18 (64)	5 (18)
Acceptance	16 (57)	9 (32)
Insecurities and worrying	12 (43)	3 (11)
Social relationships and support	12 (43)	1 (4)
Emotions and tension	10 (36)	1 (4)

Step 2: Develop and Validate Program Content and Structure

Consulting Dementia Care Experts

Methods

Individual in-depth interviews were conducted to explore dementia care experts' views on EDC Internet support. Experts from different institutions and regions within the Netherlands were recruited via email. Inclusion criteria were (1) professional caregiver in the Dutch dementia care field, (2) daily interaction with people with dementia (PwD) and their caregivers, and (3) ample experience in supporting EDC. The number of participants (N=11) was determined by data saturation. The redundancy of themes emerged from interviews [34]. Professional backgrounds of the experts were psychiatrists (N=1), clinical neuropsychologists (N=3), registered health psychologists (N=4), occupational therapists (N=1), social psychiatric nurses (N=1), and nurse practitioners (N=1), with an average of 13.64 (SD 7.43) years of professional experience. A semistructured interview guide was developed by authors LMMB and MEdV, which was validated by author FRJV. Topics included EDC needs, relevance and feasibility of EDC support, and themes for an EDC intervention. Brief summaries of the key points were made throughout the interview to obtain participant verification [35]. All interviews were audiotaped. The content of the verbatim transcriptions of the interviews was analyzed by summarizing common themes based on deductive content analysis.

Results

The experts emphasized that caregiver support needs to be tailored to the dementia stage. Concerns were raised about providing early support in the absence of later-stage problems, when caregivers are not in need of help yet and could possibly reject early stage support. Experts considered education on the disease and its course as most important. Other important themes involved accepting the disease, coping with relationship changes, stress, role management, and rumination. The importance of interaction between PwD, caregivers, and environment was also stressed. Correcting or accepting care recipients' mistakes and notifying social network can be primary stressors in daily interaction. Too much negative information not fitting the early stages, for example, behavioral problems, care homes, and end-of-life decisions, could lead to adverse reactions and should be avoided in EDC support.

Combining the themes chosen by the experts and the caregivers resulted in 9 separate themes (Table 3). The theme "practical tips" was incorporated in the module structure to provide tips thematically.

Table 3. Modules of "Partner in Balance" and their key points.

Module	Key points
Acceptance	Identify changeable and unchangeable situations
	Adapt expectations and learn to let go
Balance in activities	Change in daily and pleasant activities
	Identify personal carrying capacity and burden
Communication with family member and environment	Communication changes due to memory problems
	Effective communication with adaptations
Coping with stress	Relationship between stress and health problems
	Identify and cope with stress in daily life
Focusing on the positive	Identify activities and situations that are still possible
	Find alternatives and accept adaptations
Insecurities and rumination	Recognize rumination signals and control thoughts
	Prevent looking ahead; live in the moment
Self-understanding	Self-evaluation in caregiver encounters
	Personal strengths and areas of improvement
The changing family member	The changing memory and behavior
	Influence of memory decline on daily life together
Social relations and support	Value and maintenance of social relations
	Types of support

Content Proposal: Existing Evidence and Conceptual Frameworks on Self-Care

Methods

Intervention content was proposed by authors LMMB and MEdV based on a literature review [10], EDC needs [24], identified themes in Step 1, and conceptual frameworks on self-management. The Stress and Coping paradigm [25] served as the theoretical basis for the content of the modules. According to this model, stress is experienced when a person perceives that the demands (caring for a person with dementia) exceed their personal and social resources. Caregivers' responses to their stress situation might be mediated by their understanding of the situation and their beliefs about their ability to cope. The latter fits Bandura's [26] concept of self-efficacy (belief in one's capabilities). Consistent with this theory, models of dementia management emphasize the need to maintain self-worth and control [28]. An intervention aimed at increasing self-efficacy should not only educate the caregiver, but should also foster self-management by combining education with problem-solving skills, and work toward a change in behavior [36].

Results

The proposed self-management intervention program "Partner in Balance" (PiB) encourages caregivers to actively manage their lives and identify solutions for their specific needs [37]. Increasing knowledge, identifying and setting goals, and learning skills to achieve these previously set goals served as the basis for the intervention program. Module content was focused on role management (eg, balancing activities in daily life) and emotional management (eg, dealing with fear and insecurity about the future) [38]. Formulating, planning, and executing personal goals can be learned using a proactive 5-step change plan (Textbox 1) often used in self-management [38], which was integrated into each module. By formulating and planning a personal change plan, caregivers learn to anticipate on stressful situations and gain confidence in their ability to take care of the situation and themselves [38]. Because caregivers greatly varied in their needs, personal goals, and interest, a flexible choice of modules was used. Successful elements that were identified in the literature review [10], including tailored caregiving strategies and contact with a coach and/or other caregivers, were included in the program content likewise.

Textbox 1. The five self-management steps applied in each module.

Step 1: Recognize areas that you wish to change or to maintain

Step 2: Recognize additional conditions and barriers

Step 3: Generate alternative strategies for the problem(s)

Step 4: Write down your final plan SMART (specific, *m*easurable, *a*ttainable, *r*ealistic, and *t*imely)

Step 5: Evaluate when you will be satisfied with your progress

Validating Proposed Program Content

Methods

Dementia care experts (N=4) not involved in the initial interviews were asked to read the material and provide comments with respect to language, tone, amount of information, and significance and propriety of the content. They worked as clinical neuropsychologist (N=1), registered health psychologist (N=1), occupational therapist (N=1), and social psychiatric nurse (N=1), with an average of 9.75 (SD 2.22) years of professional experience.

Results

The experts provided comments concerning the content of each module. Stigmatizing, complex, or unclear language was reported and alternatives were provided. The textual content of the program was adapted accordingly.

Step 3: Testing the Feasibility and Preliminary Effectiveness

Think-Aloud Usability Testing

Methods

A Web-based fully operational program was developed based on the senior-friendly website checklist [39]. Initial usability flaws were tested with the "think-aloud" method [40]. Potential users were asked to think aloud while using the system, allowing the researchers to understand the reasons behind their usage behavior. Participants (N=4) were randomly selected from the focus group interviews (N=2) and the expert interviews (N=2). All aspects of the senior-friendly website guidelines were checked (eg, font size, contrast, menus and navigation, button style and size, phrasing, illustrations and videos, and Web assistance) and additional comments on user-friendliness were explored in-depth. The interviews were audiotaped and the verbatim transcripts were combined with field notes made by author LMMB on experienced difficulties during the walk-through. A coding scheme based on the interview protocol was used [40].

Results

All participants (N=4) commented on the layout, font size, contrast, tone, and navigation. The website layout was considered professional and attractive, although a uniform composition of all pages was proposed to foster cohesion. Alterable font sizes and increased contrast with the background color were suggested. Vignettes of caregivers were considered useful, but addressing people with their given name instead of their family name was considered more appealing. The tips from other caregivers were perceived as crucial. It was suggested to conclude every module with these tips to increase layout uniformity.

Piloting Feasibility

Methods

An uncontrolled pre-post-intervention pilot study with EDC was conducted to establish feasibility, as this is recommended before moving on to a larger scale effect study [31,41]. The Medical Ethics Committee of the Maastricht University Medical Centre approved this study (No NL44475.068.13, Dutch Trial registration number NTR4217). Caregivers were included in the study if they (1) were spousal caregivers of people with mild cognitive impairment [42] or mild dementia of all subtypes [43], and (2) had access to the Internet. Exclusion criteria were (1) insufficient cognitive abilities to engage in the online self-management program, (2) overburdening, (3) severe health problems (determined by the study staff), or (4) caring for PwD caused by human immunodeficiency virus, acquired brain impairment, Down syndrome, Huntington's chorea, or alcohol abuse. Participants were recruited at memory clinics and ambulatory mental health clinics. Based on comparable feasibility studies, we aimed to include 10 participants [44,45]. Of those contacted, 17 of 43 caregivers (40%) were willing to participate and signed the informed consent form.

Feasibility was evaluated face-to-face at the caregiver's home by a semistructured interview developed for this study—the Program Participation Questionnaire (PPQ). The PPQ was based on measurement scales for perceived usefulness and ease of use and overall acceptance of information technology [46,47] and included 30 items on usability, clarity, comfort with, and acceptability of the format on a 7-point Likert scale ranging from 1 (strongly disagree) to 7 (strongly agree). For the individual PPQ items, see Multimedia Appendix 1. Mean scores were calculated with descriptive statistics. Because there were no external criteria to properly define feasibility [48], we followed the conventional strategy of using the median score of the questionnaire as a cutoff. This approach was previously adopted in a Delphi study as evidence of agreement of intervention feasibility [49]. Based on the PPQ scale (range 54-234, median 144), scores of 145 or higher were considered "acceptable feasibility." Mean item scores (range 1-7) will be used to make decisions on positively and negatively appraised aspects of the program. Mean item scores of 5 (slightly agree) or higher will be considered positive, mean item score below 4 (slightly disagree or lower) will need further revisions. Participants were asked to elaborate their scores. Comments were audio recorded and transcribed verbatim. Meaningful data units based on the PPQ items were identified and derived independently from the qualitative data by authors LMMB and HEJW with deductive content analysis. Furthermore, the actual accessed data use of the program (number of log-ins and features used) was compared with self-reported data.

Results

The study population consisted of 17 participants, of whom 10 completed the postintervention assessment. Participants who did not complete the postintervention assessment were replaced to meet the sample size suggested by previous studies [44,45]. The main reasons for not completing the program and the postintervention assessment were difficulties with the online aspect of the program (N=4), private circumstances (N=2), and disagreements with the care recipient (N=1). Completer and noncompleter characteristics are listed in Table 4.

Table 4. Participant characteristics for completers and noncompleters of the intervention.

Characteristics		Completers (N=10)	Noncompleters (N=7)
Age caregiver, mean (SD)		68.10 (6.54)	67.43 (5.65)
Age care recipient (people with dementia [PwD]), mean (SD)		69.90 (4.33)	71.57 (8.46)
Hours care per week, mean (SD)		44.20 (56.85)	76.43 (71.98)
Gender, N (%)			
	Male	7 (70)	2 (29)
	Female	3 (30)	5 (71)
Education, N (%)			
	High school	1 (10)	3 (43)
	College	7 (70)	4 (57)
	Graduate school	2 (20)	0 (0)
PwD diagnosis, N (%)			
	Mild cognitive impairment	7 (70)	5 (72)
	Alzheimer's disease	3 (30)	1 (14)
	Vascular dementia	0 (0)	1 (14)

[a]Noncompleter rate=41.2%.

The PPQ showed a good internal consistency (alpha=.89) and had a mean sum score of 209 (SD 22.14). Given the threshold of 145 or higher, this score indicated an acceptable feasibility. Mean item scores above 5 (slightly agree or higher) were found for convenience of completing the program at home (5.9, SD 1.8), clarity of the website (6.0, SD 1.2), module structure (6.1, SD 0.6) and content (6.6, SD 0.5), privacy (6.6, SD 1.3), tailored assignments (6.1, SD 1.3), guidance by the coach (6.6, SD 0.5), and general contentment (6.4, SD 0.9). A mean item score below 4 (slightly disagree or lower) was found for usefulness of the discussion forum (2.8, SD 2.7). Table 5 shows the positive, negative, and neutral themes derived from the additional comments. Self-report usage data were comparable to tracked usage data: 106.41 (SD 96.15) minutes spent per module, including scoping the website (4.5 minutes, SD 4.13), completing the assignments and change plan (79.14 minutes, SD 77.16), contacting the personal coach (15.31 minutes, SD 16.96), and visiting the discussion forum (7.46 minutes, SD 7.09), spread out over 2.38 (SD 1.38) weeks.

Table 5. Positive, negative and neutral evaluation of different features of Partner in Balance (N=10).

Feature	Positive	Negative	Neutral	Quotes[a]
Online	At home Work in own time	Writing personal situations feels more confronting than verbalizing them	—	*I thought it was useful to have the ability to do it at my own convenience and only when I was in the mood for it.* [P10]
Website	Quick response during problems Fragmented information	Technical problems: login and communication		*It was pleasant that the information was presented in parts*. [P8]
Personal coach	Indispensable support Feedback boosts self-confidence and motivation	Goal setting during intake difficult	Help of coach during goal setting necessary, not able to do it alone.	*It is important that you have someone you can rely on during the program. It stimulates you even more and you know you are not alone.* [P8]
Video clips	Relatable examples Personal aspect	Not applicable to every individual Confronting	Background of caregivers unclear	*I could not identify with the video clips because it was not clear who was talking and I cannot relate to the addressed problems just yet.* [P3]
Discussion forum		Too difficult to use Purpose unclear Difficult to start conversation No nonverbal communication	Coaches could feed the forum, ask questions or outline situations	*I am not going to write something out of the blue on a discussion forum. You might say the wrong things, even if you have good intentions.* [P2]
Change plan	Useful Increases awareness	Felt like an obligation	Difficult to set personal goals	*The self-management assignment teaches you a lesson. How are you going to solve these things?* [P10]
Application in daily life	Communication tips		Already apply tips in daily life	*The coach provided me with very good tips that I applied in daily life.* [P12]
General contentment	Boosts confidence Recommend to other caregivers	No focus on practical decisions possibly faced in the future		*On moments that you are feeling insecure, this program gives you some kind of confirmation. It makes you doubt yourself less.* [P10]

[a]P (number): participants number in the feasibility study.

Piloting Preliminary Effects

Methods

Preliminary understanding of the effectiveness of the program was based on the baseline and postintervention assessment 8 weeks later, completed at the participant's own convenience in an uncontrolled pilot study. At participant's request, paper questionnaires were used. The Caregiver Self-Efficacy Scale (CSES) was used to measure domain-specific caregiver self-efficacy [50]. The subscales include 4 items on service use and 6 items on care management, with scores ranging from 1 (not at all certain) to 10 (very certain). We found a good internal consistency for both service use (alpha=.73) and care management (alpha=.87). Paired samples *t* tests were conducted to evaluate pre-post-intervention changes. The goal attainment scaling (GAS) [51] method was used to rate treatment-related change and to compare relative success of previously set personal goals. Baseline scores were set at −2. Postintervention scores can range from −2 (much lower than expected) to +2 (much better than expected), with a score of 0 meaning goal attained. Raw scores were transformed into an individual mean GAS score (*T* score) to determine goal attainment with a potential weight assigned to the goal(s) [52]. *T* scores of 50 or more (SD 10) indicate effective goal achievement.

Results

Postintervention, participants (N=10) had significantly higher scores on both the CSES care management subscale (mean 41.1, standard error [SE]=2.5; t_9=−2.5, *P*=.03) and service use subscale (mean 32.6, SE=1.7; t_9=−3.5, *P*=.01) compared with preintervention scores (mean 36.1 and SE=3.2, and mean 23.2, SE=3.4, respectively), although effect sizes were small (*d*=0.14 and 0.41, respectively).

In this study, 8 program completers set 13 goals in total; 2 program completers were not able to set goals due to personal difficulties verbalizing the desired change. Two goals (of 2 participants with multiple goals) could not be scored after the intervention, because the goals changed during the course of the study. In total, 8 goals were attained (2 attained, 5 higher than expected, and 1 much higher than expected), and 3 goals were unattained (1 much lower than expected and 1 lower than expected). The mean *T* score at baseline (set at the −2 level) was 27.8 (SD 3.04). The mean achieved *T* score after the intervention was 53.7 (SD 12.03). Table 6 shows the number

of goals for each domain, with most goals set on communication with the care recipient (N=7), followed by maintaining positive activities together (N=2), obtaining social support (N=2), and planning time alone (N=2).

Table 6. Number of set goals and attainment scores (N=8).

Number of goals	Scores	
	Mean (SD)	Range
Number of set goals per participant	1.6 (1.06)	1.0-4.0
GAS[a]score at baseline	27.8 (3.04)	22.6-30.0
GAS[a]score achieved	53.7 (12.03)	30.0-70.0

[a]GAS: goal attainment scaling

Step 4: Adapting the Program—Final Intervention

Following the iterative development process, the program was adapted according to the results obtained in the feasibility study. The discussion forum was expanded with regular posts from personal coaches with practical tips, literature, and events related to EDC. The role and background of the person in the video clips was clarified. In addition, the content of often-mentioned early stage situations and problems [24] was expanded and later-stage problems were made less prominent in the video clips. Furthermore, technical issues with logging in and communicating with the personal coach were resolved with the team of Web experts.

Final Intervention

PiB consists of three elements, namely, (1) face-to-face intake session with a personal coach, (2) online period guided by the personal coach (psychologist or psychiatric nurse with ample experience with dementia caregivers), and (3) face-to-face evaluation session with the personal coach.

Intake Session

In the intake session participants are introduced to the website and the self-management concept of the program. The coach and participant set personal goals using a motivational interviewing technique frequently used to identify change objectives and enhance intrinsic motivation [53]. Based on the discussed areas for improvement, participants select 4 of the 9 modules that were previously identified by experts and caregivers. Participants are provided with personal login codes to access their selected modules and edit their personal information. After the online period, participants will discuss their personal goals and their ability to cope with future difficulties in the evaluation session.

Online Period

During the online period of the intervention, participants follow the chosen modules during an 8-week period. The website consists of (1) a home page with a short description of the goal of the program, personal login option, contact information of the researcher, and the institutional affiliation (Maastricht University); (2) a personal page with a link to the chosen modules and a mailbox for exchanging emails with the personal coach; and (3) an online forum to interact with other caregivers, moderated by the researcher and personal coaches.

Every module has a fixed design of the following 4 components: (1) video clip of fellow family caregivers, (2) education, (3) self-reflection assignment, and (4) the 5-step change plan, guided by the personal coach who will provide individualized online feedback after completion of each module and offers assistance when needed. For every module, 2 weeks are reserved as a starting point. However, participants are allowed to complete the modules at their own pace as informed by the self-management approach [38]. The first week of a module addresses Components 1-4. Participants can send their assignment and the 5-step plan to their coach. The second week of every module is reserved for feedback from the coach, after which participants can adjust their 5-step plan if necessary.

Personal Coach

The personal coach is an experienced dementia care professional (psychologist or psychiatric nurse). Coaches will receive a 1-day training in self-management techniques and online help before the start of the intervention. They will receive experienced supervision from an experienced professional in psychology and self-management during the course of the intervention period to ensure quality and alignment of the feedback of the coaches according to self-management principle. Coaches are asked to support participants in choosing modules that fit their personal situation, help participants identify feasible goals, offer techniques to achieve goals, and provide participants with general constructive feedback on their assignments. Using a personal login code, coaches will be matched with the participants assigned to them. The CONSORT EHEALTH checklist is presented as Multimedia Appendix 2.

Discussion

In this paper, the iterative development process of the Web-based self-management program PiB for EDC was presented. Use of the MRC framework enabled us to develop an intervention based on existing research, theoretical frameworks, and user and professional input. Including potential users during the design process enabled us to gain unique insights into usage behavior and challenges to adapt the technology to the needs of the target audience. A similar design has been successfully used in previous studies [54-56].

During the exploration phase, caregivers greatly varied in their need for information. Previous self-management studies confirm that personal caregiver needs should be used as a starting point [57]. Blended care was preferred over online care only, due to

the personal contact with a professional. Previous studies support the value of this format as participants highly appreciated the connection with their coach or therapist [58] and felt more motivated to complete Web-based interventions [59]. By contrast, adding face-to-face contacts increases the costs of the online program and reducing the number of face-to-face contacts might harm treatment outcomes when online components are not used [60,61]. However, blending online modules with regular face-to-face therapy can increase the adherence and effectiveness of the treatment. Scientific validation of blended care interventions is warranted for the development and adaptation of future treatments [60] and can provide important support for the use of blended care interventions rather than online therapy only. The former can be more easily implemented by health services, therapists, and clients than online therapy, as they can be integrated into existing treatment and care settings [62].

Our results showed acceptable rates of satisfaction with PiB. Caregivers greatly appreciated the use of online resources due to the convenience of completing the program from their homes, which is in line with previous studies on Web-based caregiver interventions [46,63-65]. Furthermore, PiB supported the participants in the process of caregiving and boosted their self-confidence, probably due to the combination of support, self-management, and a tailored approach [66,67]. However, the noncompleter rate was high due to initial technical difficulties, which were resolved later on in the study. Unfamiliarity with using the website also caused difficulties among the older age group, resulting in a relatively young sample. A recent study confirmed that younger dementia caregivers were more likely to use the Internet for health-related purposes [68]. However, other research showed that homebound older adults with limited computer skills who receive computer training at the start of an intervention can participate without difficulties [69]. In addition, including more potential users in the thinking-aloud procedure could be helpful [40].

The discussion forum was negatively reviewed and hardly used, due to the unclear purpose, the anonymity of participants, and the high threshold for starting a conversation. However, other studies suggest that forums can serve as a valuable addition to share experiences and support [39,45,70]. Adding new tips or developments for caregivers could increase the use of a forum because the aspect of reading posts was already considered useful [70]. Furthermore, participants reported struggles with goal setting. This could be due to the relatively lower objective burden of EDC, compared with caregivers of people in the later stages of the disease. EDC might experience more subjective burden, which is more difficult to translate into specific needs. Problems with accepting and adapting to their new role may also have hindered goal setting [24]. Therefore, goal setting for EDC should focus on the enhancement of positive overall experiences and facilitation of the personal adaptation process, rather than exclusively aiming for change. This approach may help to reduce or prevent negative consequences of caregiving (eg, overburdening) at a later stage [24].

Preliminary effects on caregiver self-efficacy and goal attainment were small, yet positive. This finding is in line with Bandura's theory on self-efficacy, which states that caregivers' objective understanding of the situation and belief in one's capabilities (ie, the self-efficacy level) can increase if provided with the right tools [26]. Furthermore, this finding is congruent with previous research on online support for dementia caregivers [21,71,72].

The program in this study was evaluated in a homogenous group of primary caregivers (eg, spousal caregivers), with specific attention for the spousal relationship. However, the themes may apply to a broader target group, as demonstrated by previous studies [13-19]. PiB could potentially be suitable for other primary carers, which should be further investigated in the upcoming effect study using a larger sample.

Limitations

The small sample size, the lack of a control condition, and a possible sampling bias based on caregivers with access to the Internet make it difficult to generalize the results. However, previous studies adopting a development and feasibility approach have used similar methodology and sample sizes [15,73], fitting the purpose of formative research [31]. In addition, users and experts were closely involved in the development of the intervention, and the content and adaptations relied on in-depth participant and expert feedback. Future research should consider inclusion of caregivers in the proposed content validation, to ensure potential user feedback in every step of the development. Furthermore, drawing conclusions from the adopted median cutoff score, which is an arbitrary value, may not be justified. However, in this study, the overall feasibility score was not leading. Mean item scores were used to make program improvements.

This study used paper questionnaires. Although seniors' use of the Internet is expected to increase over time [74], dementia caregivers seem to be less active in health-related Internet use compared with the population at large [68]. The high noncompleter rate resulted in missing post-test data from the noncompleters as these were collected after the last module. However, reasons for noncompletion and characteristics of noncompleters were provided, giving insight into their possible motives. Future effectiveness studies should include noncompleter data after the treatment and at follow-up and consider using Web-based questionnaires, with the advantages of low costs, no missing data or data entry errors, and time flexibility [75].

Conclusion

Our tailored intervention approach appeared to be feasible for informal EDC and provided them with important support when dealing with the difficulties of caregiving. Feasibility results were used to improve the intervention. Confirmation of the feasibility and preliminary effectiveness is a valuable step toward examining the effectiveness of this intervention, as recommended by the MRC framework [31]. The PiB course is currently (November 2015) available for caregivers who are interested in participating in the effectiveness study. At the course website (Partner in Balans) they can express their interest by emailing the researcher, after which they will receive additional information about the course and the effectiveness study.

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Authors' Contributions

LMMB, MEdV, GIJMK, and FRJV designed the study. LMMB collected the data, performed the analyses and wrote the manuscript. MEdV and HEJW performed the second independent qualitative analysis for different stages in the development. MEdV supervised the data collection and analysis. MEdV, HEJW, GIJMK, and FRJV reviewed and approved this manuscript.

Conflicts of Interest

None declared.

Multimedia Appendix 1

Program Participation Questionnaire.

[PDF File (Adobe PDF File), 127KB - resprot_v5i1e33_app1.pdf]

Multimedia Appendix 2

CONSORT-EHEALTH checklist V1.6.2 [76].

[PDF File (Adobe PDF File), 761KB - jmir_v5i1e33_app2.pdf]

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Abbreviations

CSES: Caregiver Self-Efficacy Scale
EDC: early stage dementia caregivers
GAS: goal attainment scaling
MRC: Medical Research Council
PiB: Partner in Balance
PPQ: Program Participation Questionnaire
PwD: people with dementia
SE: standard error

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Original Paper

Download Your Doctor: Implementation of a Digitally Mediated Personal Physician Presence to Enhance Patient Engagement With a Health-Promoting Internet Application

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Abstract

Background: Brief interventions delivered in primary health care are effective in reducing excessive drinking; online behavior-changing technique interventions may be helpful. Physicians may actively encourage the use of such interventions by helping patients access selected websites (a process known as “facilitated access”). Although the therapeutic working alliance plays a significant role in the achievement of positive outcomes in face-to-face psychotherapy and its development has been shown to be feasible online, little research has been done on its impact on brief interventions. Strengthening patients’ perception of their physician’s endorsement of a website could facilitate the development of an effective alliance between the patient and the app.

Objective: We describe the implementation of a digitally mediated personal physician presence to enhance patient engagement with an alcohol-reduction website as part of the experimental online intervention in a noninferiority randomized controlled trial. We also report the feedback of the users on the module.

Methods: The Download Your Doctor module was created to simulate the personal physician presence for an alcohol-reduction website that was developed for the EFAR-FVG trial conducted in the Italian region of Friuli-Venezia-Giulia. The module was designed to enhance therapeutic alliance and thus improve outcomes in the intervention group (facilitated access to the website). Participating general and family practitioners could customize messages and visual elements and upload a personal photo, signature, and video recordings. To assess the perceptions and attitudes of the physicians, a semistructured interview was carried out 3 months after the start of the trial. Participating patients were invited to respond to a short online questionnaire 12 months following recruitment to investigate their evaluation of their online experiences.

Results: Nearly three-quarters (23/32, 72%) of the physicians interviewed chose to customize the contents of the interaction with their patients using the provided features and acknowledged the ease of use of the online tools. The majority of physicians (21/32, 57%) customized at least the introductory photo and video. Barriers to usage among those who did not customize the contents were time restrictions, privacy concerns, difficulties in using the tools, and considering the approach not useful. Over half (341/620, 55.0%) of participating patients completed the optional questionnaire. Many of them (240/341, 70.4%) recalled

having noticed the personalized elements of their physicians, and the majority of those (208/240, 86.7%) reacted positively, considering the personalization to be of either high or the highest importance.

Conclusions: The use of a digitally mediated personal physician presence online was both feasible and welcomed by both patients and physicians. Training of the physicians seems to be a key factor in addressing perceived barriers to usage. Further research is recommended to study the mechanisms behind this approach and its impact.

Trial Registration: Clinicaltrials.gov NCT 01638338; https://clinicaltrials.gov/ct2/show/NCT01638338 (Archived by WebCite at http://www.webcitation.org/6f0JLZMtq)

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KEYWORDS

alcohol drinking; physician-patient relations; behavior and behavior mechanisms; research design; Internet; multimedia; primary health care; family physicians

Introduction

Alcohol is the third leading cause of diseases and premature death globally [1], and it has been estimated that it accounts for 3.8% of deaths and 4.6% of disability-adjusted life years [2]. Brief interventions delivered in primary health care settings have been shown to be both efficacious and effective in reducing excessive drinking in Italy and worldwide [3-7]. However, some individual large-scale pragmatic trials have failed to report significant findings [8-10], and no reductions in drinking or very small effects were observed in results from studies in probation offices, emergency departments, and among college students in Sweden and New Zealand [11-14]. Remarkably, the optimal contents and delivery strategies and the impact of the brief interventions on alcohol problems and specific population groups are not clear [7]. Comparing different brief intervention programs may be challenging because of differences in their contexts, the implementation strategies, and the indicators employed [6]. Moreover, there are important barriers to implementation, such as insufficient training, lack of resources, and time constraints, which prevent brief interventions from being employed widely [4,15-17].

Internet interventions based on behavior change techniques may be helpful to tackle these barriers [4,18]. Health care professionals may actively encourage the use of such digital applications by helping patients access selected websites (this process is known as “facilitated access”) [19]. Initially adopted primarily for the management of patients with mental health problems including depression and anxiety [20], facilitated access has been extended to addictive behaviors including smoking cessation and alcohol screening, as well health promotion and the management of long-term conditions [4,21-25]. Ensuring effective patient interaction with the website following facilitated access may be an issue [26], and additional mechanisms are therefore likely to be necessary to achieve more consistent engagement. In this paper, we describe the development and use of digitally mediated personal physician presence to enhance patient engagement with an alcohol-reduction website as part of the experimental online intervention in a noninferiority randomized controlled trial [27]. The intervention was designed to reinforce patients’ perception of their physician’s endorsement of the website and thus promote the development of an effective therapeutic working alliance between the patient and the application.

Described in a seminal paper by Bordin [28], the therapeutic working alliance refers to the personal relationship between the therapist and patient that stems from a collaborative endeavor to attain the goals of the treatment. Such an agreement on goals and tasks and the establishment of a personal bond of reciprocal positive feelings constitute the cornerstones of the alliance. There is a large body of evidence suggesting that it plays an important role in the achievement of positive outcomes in face-to-face psychotherapy [29-31], where patients who experience a strong alliance show more motivation and invest more effort to complete their treatment [30]. The importance of the alliance is also highlighted by researchers who suggest that nonspecific factors are largely responsible for the outcomes of different psychotherapies; these factors are common in the majority of the therapeutic interventions and include the healing setting, the expectations of improvement, and the therapeutic bond [32]. Little research has been done on the role of the therapeutic working alliance in treatment of alcohol problems or in brief interventions, but evidence is beginning to emerge that it may have an important influence on patient outcomes [33].

Researchers argue that the quality of the therapeutic working alliance is largely determined by the therapist variation, which may also explain the therapist’s effects on the outcomes [32]. Patient expectations of Internet-delivered behavioral change techniques may play a significant role as well [31], and according to Jasper et al, the development of the therapeutic alliance online may require more time than when therapy is delivered face-to-face [34]. In the context of facilitated access, agreement on goals and tasks can generally be achieved relatively easily online through the offer of a menu of options [34,35], but the maintenance and enhancement of the therapeutic alliance is considerably more problematic.

In health-promoting websites selected for facilitated access, customization of the information delivered to patients combined with multimedia emulation of their physician’s presence online may increase patient engagement, potentially by strengthening the online therapeutic alliance. Based on this hypothesis, we developed the digitally mediated primary care physician presence (Download Your Doctor) as a mechanism for achieving this end. We describe below how this was implemented in the context of a trial designed to determine whether facilitated access by primary care physicians to an alcohol-reduction

website was superior to traditional face-to-face brief intervention [27] and to report the feedback of the users on the module.

Methods

The Download Your Doctor module was developed as an additional feature to an alcohol-reduction website [36] with the objective of generating a simulated personal physician presence in the online environment. The website was developed as part of a randomized controlled trial conducted in primary health care settings in the Italian Region of Friuli-Venezia-Giulia (EFAR-FVG), aiming to test the effectiveness of facilitated access to an alcohol-reduction website compared with standard face-to-face brief intervention [27,36]. Its contents and design were based on the UK counterpart, DownYourDrink, which has been described elsewhere [37,38]. The rationale for the trial is that general practitioners (GPs)/family physicians (FPs) are generally reluctant to screen for alcohol misuse and delivery rates of brief interventions are low [27,39]. The use of facilitated access to a selected website to provide patients with an opportunity to undertake screening and brief intervention online rather than in the consultation offers significant promise, but a trial was needed to establish noninferiority of this approach. The study protocol was approved by the Isontina Independent Local Health Unit Ethics Committee on June 14, 2012.

For the duration of the trial, the participating GPs/FPs provided facilitated access consisting of a short discussion followed by presentation to the patient of a pamphlet describing the website and containing the patient's personal login code. The first 3 digits of the login code were specific to the general practitioner and the last 4 to the patient. The online alcohol screening module was based on the 3-item Alcohol Use Disorders Identification Test (AUDIT-C). Those scoring above the designated cut point were requested to provide consent to the trial and then undergo an online baseline assessment, followed by the randomized assignment to receive either online facilitated access to the alcohol-reduction website or the face-to-face intervention by their GP/FP. Follow-up assessment took place online at 3 and 12 months using the full version of the AUDIT questionnaire [40] and the EQ-5D [41].

The Download Your Doctor facility enabled the GPs/FPs participating in the study to personally customize selected messages and visual elements of the user interface to be presented to their patients after logging on to the website and following screening (see Figure 1). Each practitioner was additionally able to upload their personal photograph and signature for integration with their messages on the website. The appearance of each message could thus be customized by inserting links and images shown in a "speech bubble" next to text and along with the photo of the GP/FP (see Figures 2-4). Finally, they were also given the option to upload video recordings in order to simulate online communication with their patients even more directly.

Before the onset of the trial, the participating GPs/FPs were invited to attend a training session, during which a stepwise presentation of the website was carried out and the Download Your Doctor module was explained with interactive examples. Particular attention was given to the ways the physicians could enhance patient participation and adherence to the study. Subsequent use of the module by each participating physician was recorded by the study team.

To assess the perceptions and the attitudes of the physicians with regard to their participation in the study, an interview was conducted 3 months after the start of the trial. The interview was conducted by 2 psychologists using a semistructured questionnaire format. It sought information on such aspects of the trial as the nature of physicians' interactions with the patients (recruitment and instructions) and their satisfaction with the participation in the trial. During the interview, the use and evaluation of the website (personalization, usability, and usefulness of the content) were also explored.

Moreover, a short online questionnaire was developed post hoc by the research team to evaluate the perceptions and attitudes of the trial participants about their experience in the study, their use of Internet and health-related websites, and the quality of their online experiences. A number of questions specifically sought information about the patient perceptions of the importance of the features related to the Download Your Doctor module. Five categories were available to rate the features ("highest," "high," "average," "low," "lowest"). Completion of the questionnaire was optional, and it was included in the final assessment that each patient was requested to undertake online 12 months following recruitment to the trial.

Figure 1. The GP/FP customization page (adapted in English).

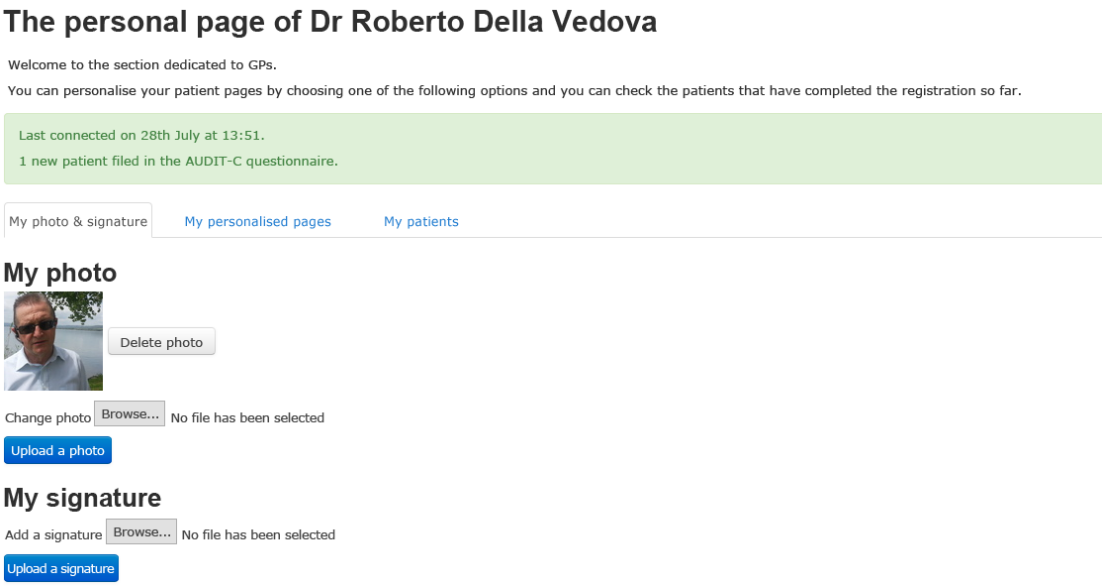


Figure 2. The screening page with the personalized speech bubble (adapted in English).

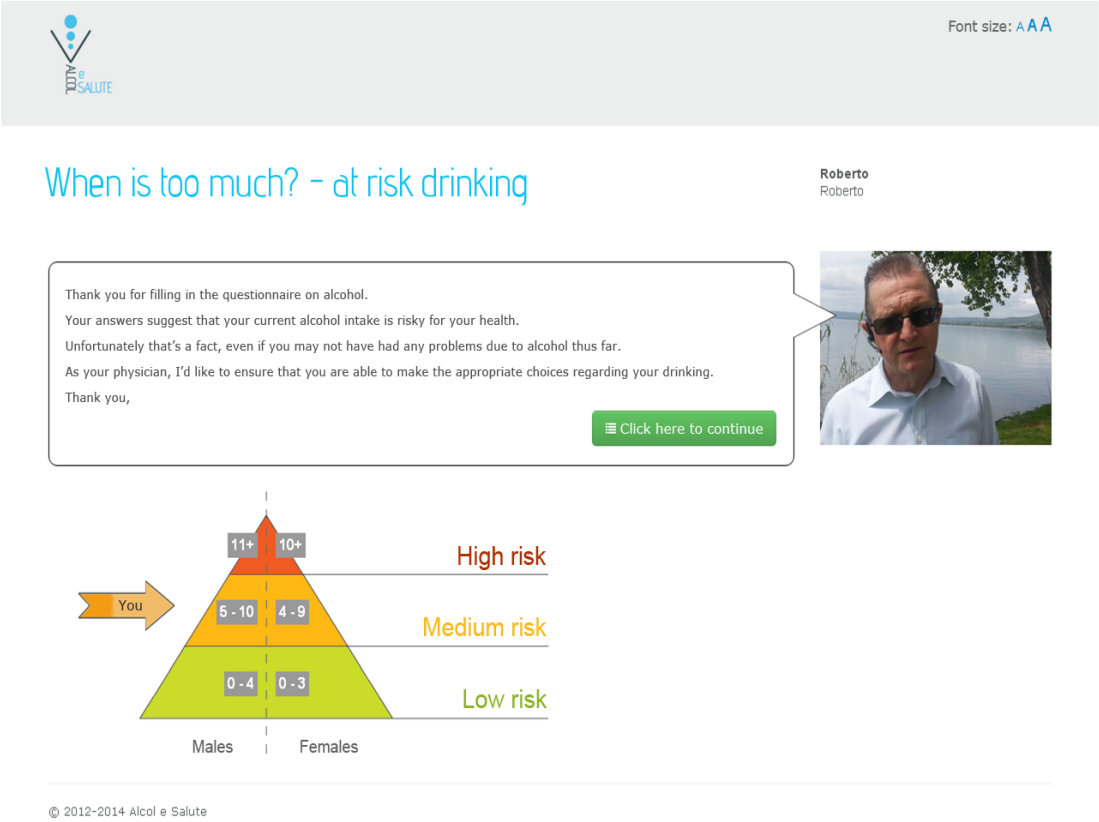
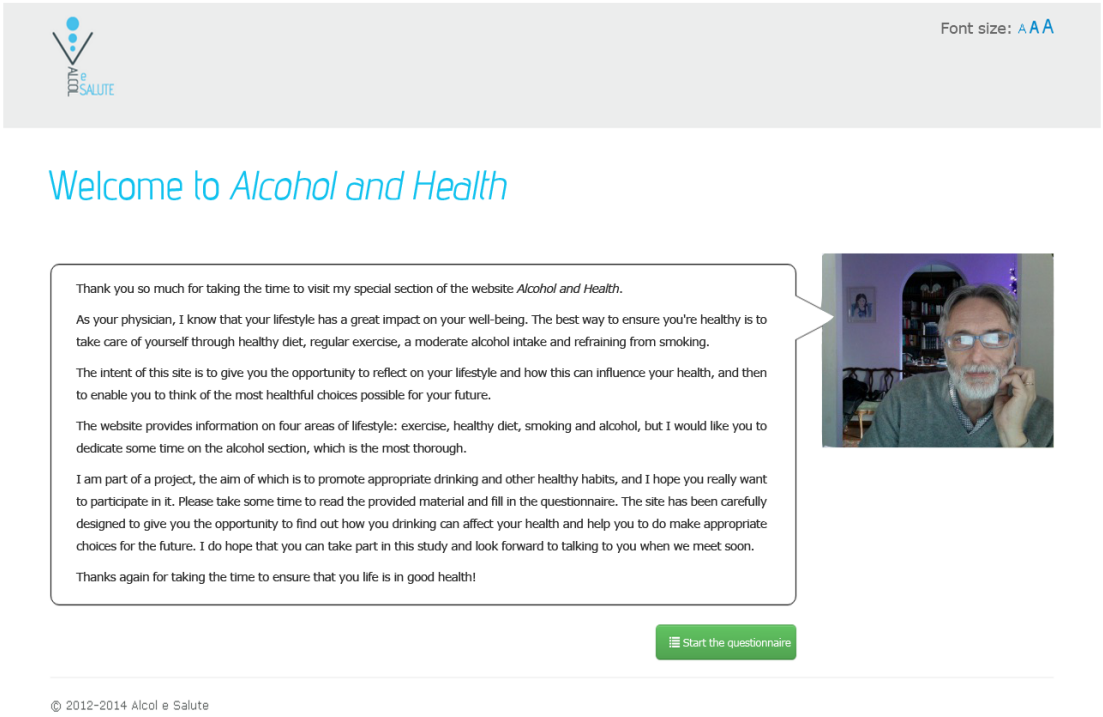


Figure 3. The personalized welcome page (adapted in English).



Results

Of the 44 participating GP/FPs approached by the research team, 32 (73%) agreed to be interviewed, and 23 (72%) of those used the Download Your Doctor module and personalized some of its parts by uploading photos and/or adding their own text messages (see Table 1).

Table 1. Personalization of the website (and lack of) among interviewed doctors (N=32).

Personalization details	n (%)
Interviewed doctors who personalized one or more elements of the website	23 (72)
The introductory photo and video	21 (57)
The introductory text	11 (34)
Interviewed doctors' reasons for not personalizing the website	9 (28)
Limited time	3 (33)
Seemed to be complicated	2 (22)
Seemed not useful	2 (22)
Unwilling to publish personal photos on the Internet	3 (33)

Physicians' Evaluation of the Website

According to the feedback from the physicians, the website scored highly in the aspects of user-friendliness and intuitive navigation, while overall the contents were considered to be useful for their patients (see Table 2). No significant differences emerged between physicians who personalized the website and those who did not do so with regard to the aforementioned aspects of user-friendliness and content usefulness, as well as the level of interest and overall satisfaction with the participation in the trial (P>.10).

Table 2. Physician feedback on the website based on Likert scales, ranging from 1-10 (lowest-highest).

Survey question	Median	Interquartile range
To what degree do you find the website easy to browse and interact with?	10	8-10
To what extent do you rate the website's contents and activities to be useful for the patients?	8	8-10

Patients' Evaluation of the Website

Of the 620 patients who completed the 12-month follow-up questionnaires, 341 (55.0%) also completed the optional questionnaire. The demographic characteristics and information technology (IT) skills of the sample are shown in Table 3.

Figure 4. The GP/FP can provide customized comments on the different patient inputs (adapted in English).

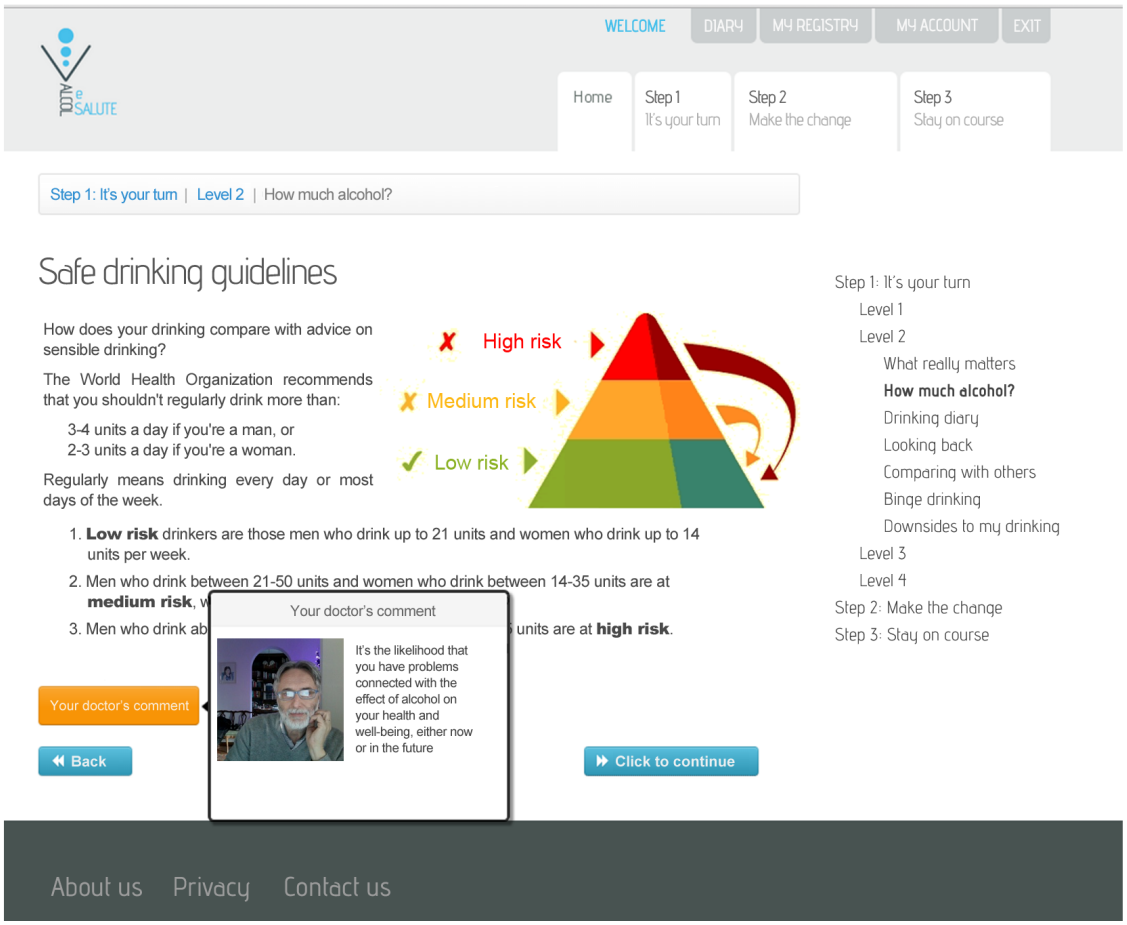


Table 3. Demographic characteristics and IT skills of the sample (N=341; only valid answers are shown).

Characteristics	Values
Gender, n (%)	
Males	215 (64.2)
Females	120 (35.8)
Age, mean (SD) interquartile range	47.27 (14.60) 36.00-58.00
IT skills, n (%)	
Poor	47 (14.0)
Basic	90 (26.9)
Intermediate	82 (24.5)
Advanced	116 (34.6)

Nearly three-quarters (240/341, 70.4%) of the patients recalled having noticed the personalized elements of their GPs/ FPs (see Table 4). There were no significant differences in the gender, age, and IT skills between the participants who noticed the

personalization and those who did not (*P*≥.05). Similarly, the two groups did not differ in the frequency with which the participants used the Internet and various health-related websites (*P*≥.05).

Of those patients who noticed the personalization, the majority (208/240, 86.7%) considered such an approach to be of either high or the highest importance for their level of confidence towards the information provided by the website (see Table 4). There were no significant correlations between the level of their confidence and the age, IT skills, and frequency with which the Internet and the health-related websites were employed (*P*≥.05), while the level of their confidence did not differ between the two sexes (*P*≥.05).

Furthermore, approximately half of those who did not notice any personalized element (55/101, 54.4%) considered such a feature to be potentially of either high or the highest importance (see Table 4).

Table 4. Patient feedback on website (N=341).

Questions	n (%)
All respondents (n=341): "When you first logged into the website, do you remember having seen a photo or a text message from your GP?"	
Yes	240 (70.4)
No	101 (29.6)
Respondents who recalled GP personalization of the website (n=240): "To what extent do you think that seeing a photo or a message from your GP improved your confidence towards the information provided by the website?"	
Lowest	1 (0.4)
Low	5 (2.1)
Average	25 (10.5)
High	74 (31.0)
Highest	134 (56.1)
Respondents who did not recall GP personalization of the website (n=101, valid answers=100): "To what extent do you think that seeing a photo or a message from your GP could have improved your confidence towards the information provided by the website?"	
Lowest	15 (14.9)
Low	14 (13.9)
Average	17 (16.8)
High	37 (36.6)
Highest	18 (17.8)

With regard to the participants' evaluation of the website, only a few replies differed significantly between those who noticed the personalization and those who did not recall such elements (see Table 5). There were no significant differences between the two groups in the subjective evaluation of the individual alcohol consumption and the role that the website played (*P*≥.05): "Do you think that you have reduced your alcohol intake compared with a year ago?," "Do you consider that your way of drinking today poses a risk to your health?," "Do you think that the website was helpful in changing your way of drinking alcohol?". Finally, the majority of the patients considered the website very easy to use, and no differences were found between the two groups (median 4.52 for those who recalled the personalization vs median 4.35 for those who did not recall such elements).

Table 5. Significant differences in the replies between the participants that recalled the personalization and those who did not do so. Answers were based on a 5-point Likert scale, ranging from 1-5 (lowest-highest).

Questions	N	Mean	Standard deviation	*t* (df)	*P*
"Do you assess your participation in the study positively?"					
Respondents who recalled the personalization	238	4.09	0.86	3.18 (317)	.002
Respondents who did not recall the personalization	101	3.74	1.04		
"Did you find the contents of the site interesting?"					
Respondents who recalled the personalization	108	4.31	0.76	2.85 (154)	.005
Respondents who did not recall the personalization	48	3.90	0.97		
"Would you recommend the website to others?"					
Respondents who recalled the personalization	108	4.23	0.82	2.34 (154)	.020
Respondents who did not recall the personalization	48	3.88	1.00		

Discussion

Principal Findings

The Download Your Doctor module was developed to enhance the therapeutic alliance between patients and physicians, in order to improve the outcomes of a trial on facilitated access to an alcohol-reduction website. Our hypothesis was that while patients may appreciate the opportunities presented through facilitated access, there might be a risk that they perceive it as less valuable than face-to-face intervention. It was therefore important to provide for continuity of the therapeutic alliance while the patient was online. The GPs/FPs were provided with features enabling them to customize the contents of the interaction with their patients and the majority of them chose to do so, acknowledging the ease of use of the online tools. Most patients reported noticing the novelty of this approach and reacted positively to such customizations.

Providing general health communication materials may have limited effect when the information is not relevant to the individual's context [42]. Tailored communication has been used in behavioral interventions delivered in print and by telephone. Yet the advances of technology have yielded new possibilities for this increasingly common practice [43], and interventions using computer tailoring to assess individuals and deliver adapted content and feedback according to the user's characteristics have been proven to result in more behavioral changes compared to standard programs [42,44,45]. Such interventions may be more effective as they contain fewer unnecessary messages and increase the relevance of the information to the individuals, who are more likely to assimilate it [43-48]. Although interventions using social networks seem to be effective in behavioral change and higher recruitment and retention may be achieved by taking advantage of the participants' existing social networks [49], their implementation in alcohol-reduction applications seems to be scarce [18]. Furthermore, personalized feedback based on the patient's answers may have a positive influence on the therapeutic working alliance in Internet-delivered programs [18,30] and there is some evidence that the use of multimedia emulating a human therapist may further enhance its strength [50,51]. Self-help texts may also help foster an alliance based on the assumption that the patient attributes the preparation of the text to an empathetic clinician thus considering him/her present during the program [35,52].

As with many digital health applications, barriers to usage may include time limitations, insufficient skills, and privacy concerns [53-55]. The level of digital literacy of the practitioners may have played a role in whether they customized the contents of the module or not. Even though digital literacy was not assessed, the feedback from physicians relating to navigation and interaction with the website was generally positive and suggested that it was feasible for the majority to carry out the tasks of personalization after the training session. In fact, training seems to be a key factor in addressing the perceived barriers to usage, and according to literature it can also tackle the time constraint concerns, as more physicians are able to recognize the potential usefulness of the proposed tool [55].

The study was exploratory in nature and has a number of limitations. Notwithstanding the patients attributing a high significance to the customization, its impact on the outcomes of the trial could not be robustly evaluated due primarily to the variable degree of customization applied by the GPs/FPs. Not only did the content and multimedia elements vary among the participating physicians, but also some of them delayed customization until after the onset of the study. The main trial was designed to measure the effectiveness of the facilitated access to a dedicated alcohol-reduction website compared with the standard face-to-face brief intervention in primary care settings, while the evaluation of the Download Your Doctor module was added post hoc. As a result, the methodology and the available data were not adequate to enable us to draw firm conclusions. For example, we were unable to exclude "false positive" cases, namely patients who stated that they had noticed customization elements when their doctors had not actually made any modifications to the original presentation of the website. Nonetheless, while the study should be considered exploratory, as far as we are aware it is the first to address the impact of a digitally mediated primary care physician presence in an online application. As such, we have reported primarily on the feasibility of the implementation of this approach, which provides a foundation for future research into improving the quality of online therapeutic alliances by offering continuity of the already established relationship between patient and physician in the context of facilitated access to an intervention website.

Conclusion

We have described a novel approach using a digitally mediated primary care physician presence based on customized content by GPs/FPs in order to increase patient engagement and promote their online therapeutic working alliance. Our findings suggest that this approach was both feasible and welcomed by both patients and physicians. As behavior change interventions are based on multiple techniques that may work synergistically and have variable effects in different patient groups [18], further investigation should focus on determining the mechanisms of this approach and its impact on the outcomes facilitated access to Internet-delivered brief interventions programs.

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Conflicts of Interest

CL is the cofounder and Chief Executive Officer at Lumos Medica Srl, which provides software solutions for clinical trials. PW has intellectual property rights for the Downyourdrink website, is Chief Medical Advisor to the UK charity Drinkaware, and has provided private consultancy on the topic of screening and brief interventions to several agencies.

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Abbreviations

AUDIT: Alcohol Use Disorders Identification Test
EFAR-FVG: effectiveness of primary care based Facilitated access to Alcohol Reduction website – a randomized controlled noninferiority trial in Region Friuli Venezia Giulia, IT
GP: general practitioner
FP: family physician
IT: information technology

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Original Paper

Bariatric Surgery for Morbid Obesity: Tehran Obesity Treatment Study (TOTS) Rationale and Study Design

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Abstract

Background: Obesity is a major health concern in the Middle East and worldwide. It is among the leading causes of morbidity, mortality, health care utilization, and costs. With bariatric surgery proving to be a more effective treatment option for overweight and obesity, the need for systematic assessment of different procedures and their outcomes becomes necessary. These procedures have not yet been described in detail in our region.

Objective: We aim to undertake a prospective study evaluating and comparing several surgical bariatric procedures in an Iranian population of morbid obese patients presenting to a specialized bariatric center.

Methods: In order to facilitate and accelerate understanding of obesity and its complications, the Tehran Obesity Treatment Study (TOTS) was planned and developed. This study is a longitudinal prospective cohort study in consecutive patients undergoing bariatric surgery. TOTS investigators use standardized definitions, high-fidelity data collection system, and validated instruments to gather data preoperatively, at the time of surgery, postoperatively, and in longer-term follow-up.

Results: This study has recruited 1050 participants as of September 2015 and is ongoing.

Conclusions: This study will ensure creation of high-level evidence to enable clinicians to make meaningful evidence-based decisions for patient evaluation, selection for surgery, and follow-up care.

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KEYWORDS

obesity; overweight; weight loss; bariatric surgery

Introduction

Health Burden of Severe Obesity

Obesity is a growing health concern in Iran and is now a global pandemic. The latest World Health Organization (WHO) report on obesity indicates that the overweight and obese population is growing. In fact, the prevalence of obesity has nearly doubled worldwide since 1980, and more than 10% of the world's adult population is obese [1]. Although the obesity prevalence remained unchanged during the last 10 years in the United States [2], it is still high: 35.5% of the adult population in the United States is obese, which is defined as a body mass index (BMI) ≥30, 15.5% have a BMI over 35, and 6.3% are morbidly obese (BMI ≥40) [3]. Obesity prevalence is on the rise in developing countries due to demographic, socioeconomic, and nutritional transitions [4], and Iran is no exception: 10.8% and 3.4% of the population are obese and morbidly obese, respectively [5].

Severe obesity is associated with comorbidities such as type 2 diabetes mellitus, hypertension, cardiovascular disease, degenerative joint disease, and sleep apnea [6,7]. It has a major impact on quality of life [8] and psychosocial health as well, and major depression is seen in 7% of this population [9].

Treatment of Severe Obesity

Over the past few decades, there has been a major change in trends of obesity treatment. Lifestyle modifications can, at best, induce a 5-10% weight loss and improve obesity-related morbidities to a limited extent [10]. However, advancements of bariatric surgery in less invasive and safer techniques, along with the evidence-supported superior results over lifestyle modifications, rendered surgery a better treatment option [11-13]. A recent meta-analysis found a 26 kg weight difference between surgical versus non-surgical treatment of morbidly obese patients in 1-2 years follow-up [14]. A Utah obesity study found this difference at 27% of initial body weight between the two groups after 6 years [15]. Results of the Swedish Obese Subjects study after 20 years of follow-up showed 18% mean weight loss for the surgical group, compared with 1% for the non-surgical [16]. Another meta-analysis including 161,756 patients showed BMI loss of 12-17 kg/m^2 after 5 years following bariatric surgery [17]. Overall, evidence now supports the choice of bariatric surgery over lifestyle changes for the treatment of severe obesity not only because of excess weight loss (EWL) reductions, but also because of significant benefits in terms of comorbidities and prolonged survival [13,18,19]. However, surgery is not without risks; perioperative mortality is estimated about 0.3%, and 30-day complication rate about 4.1% [20].

Results of Bariatric Surgical Procedures

There are various surgical procedures for the treatment of severe obesity. Roux-en-Y gastric bypass (RYGB), laparoscopic adjustable gastric banding (LAGB), laparoscopic sleeve gastrectomy (LSG), and biliopancreatic diversion with or without duodenal switch (BPD/BPD-DS) are the recommended and most commonly performed today [21]. Laparoscopic procedures are in general preferred over the open approach [11]. Newer techniques such as plication and mini-gastric or single loop gastric bypass are also emerging. Besides the variable effectiveness across procedures, each has risks and benefits.

In terms of effectiveness, since no single study has incorporated all the techniques, inference must be made through pooled data analyses. In general, BPD/BPD-DS results in more EWL (especially for patients with a very high BMI). RYGB and LSG have shown somewhat comparable results, although some studies have shown a significant advantage for RYGB [22]. LAGB comes after these techniques. Their effect on improvement of comorbidities follows the same order [23,24].

In terms of complication rate and safety, some studies have shown a significant difference between procedures. A meta-analysis of 85,048 patients found a 30-day mortality of 1.11% for BPD-DS, 0.16% for RYGB, and 0.06% for AGB [25]. Another study evaluating LSG found its rate between LAGB and RYGB to be 0.11%, 0.05%, and 0.14% respectively [22]. Overall complication rate is estimated between 4-25% [26]. Most common adverse effects after bariatric surgery are iron deficiency anemia after bypass operations (15%) and reoperations (8%) [14]. RYGB, among others, is associated with higher complication rates, while LAGB showed more reoperation rates, up to 35% according to a large cohort study [27].

There is still no universal agreement to recommend one procedure over another. Choice of the procedure depends on many factors such as the available expertise, risk stratification, patient preferences, and goal of therapy (weight loss vs glycemic control) [25].

Knowledge Gap

We aim to undertake a prospective study evaluating and comparing several surgical bariatric procedures in an Iranian population of morbidly obese patients presenting to a specialized bariatric center. As newer techniques such as LSG (as a stand-alone operation) and mini-gastric bypass are emerging and gaining popularity, their role in bariatric surgery needs to be evaluated before incorporation into general practice.

This is one of the very first studies of its kind in the Middle East and addresses the knowledge gap on the effectiveness,

safety, and efficacy of bariatric surgical techniques, including LSG, mini-bypass, and gastric plication [28].

Methods

Objectives and Study Variables

The main objectives of the study are to identify perioperative problems of morbidly obese patients; to assess and compare the effectiveness of different bariatric surgical techniques on metabolic syndrome and other obesity-related comorbidity, by measuring anthropometric indices, EWL rates, glucose homeostasis, blood pressure, lipid profile, hormone levels, and nutritional status; to assess the psychosocial aspects of obesity before and after the bariatric surgery, and long term, including quality of life and depression; and to explore the mechanisms and underlying pathophysiology in the field of obesity and its comorbidities through pathological, genetic, and molecular studies. The study variables are summarized in Table 1.

Overall Study Design

Tehran Obesity Treatment Study (TOTS) is an ongoing, single-institution, prospective study commencing March 2013. The TOTS enrolls patients to undergo a bariatric procedure based on an individualized clinical decision plan. It is organized into 4 phases: preoperative evaluation, operation, short-term follow-up, and long-term follow-up.

Preoperative Evaluation

Baseline data collected by the research team includes demographic data, anthropometric indices, physical examination, quality of life score, psychological data, and physical activity levels. Patients are then referred for several assessments including cardiac and respiratory (including ECG, echocardiogram, chest x-ray, pulmonary function test, and/or polysomnography), gastrointestinal (endoscopy and/or barium meal and abdominal ultrasound), endocrine, and psychological assessments. Patient participation depends on approval by all the consultants. Blood and urine samples are collected preoperatively (see Multimedia Appendix 1). An obesity expert physician then assesses each individual's data. Written informed consent is obtained from all the participants, including minors and adolescents who are told fully about the study. After their approval for participation, written consent is obtained from their parents or guardians.

Operation

Patients undergo one of several different bariatric procedures. Detailed data regarding anesthesiology, operation, and recovery are collected.

Short-Term Follow-Up

Post-operative follow-up for complications of surgery as well as other issues are sought and documented. Data regarding anthropometric indices and physical examination are collected at 1 month and 3 months after surgery. At the next visit 6 months postoperatively, a more detailed assessment, including blood samples, is done.

Long-Term Follow-Up

Patients are followed annually and reassessed on all baseline variables.

Subject Recruitment and Eligibility For Surgery

Severely obese patients presenting to the Tehran Obesity Treatment Center are examined by an obesity expert in the clinic to evaluate whether they meet the study inclusion criteria. Patients then attend a free-of-charge monthly comprehensive seminar in order to increase their awareness on the subject and promote active participation in the treatment plan. After providing written informed consent, patients proceed with individualized comprehensive sessions and decisions about suitability of surgery and the specific technique are made.

Inclusion criteria were 15-65 years old, BMI levels ≥40 kg/m^2 or 30<BMI<35 kg/m^2 with a medical comorbidity/failure of intensive medical treatment for at least 1 year, acceptable surgical risk, and able and willing to provide informed consent and assure regular follow-up.

Exclusion criteria were obesity due to a treatable medical disease (eg, endocrine abnormality); any other medical, psychological, or social condition which, in the opinion of the investigators, would interfere with safe completion of the study protocol; high operative risk; contraindication to bariatric surgery or weight loss; active drug addiction; nursing, pregnant, or intending to become pregnant in the following year; and unable or unwilling to complete questionnaires or expected to experience difficulty with attendance of visits or completion of study.

Surgical Procedures

Bariatric procedures that are performed in this study include RYGB, LAGB, LSG, and mini-gastric bypass. Newer techniques such as gastric plication are also considered as a treatment option in suitable candidates. In order to record all aspects of the procedures from pre-op to postop, we broke down each of the surgical procedures into its components (eg, length of alimentary limb, pouch size) and structured a measuring scheme.

A single surgical team will perform all operations under general anesthesia, with the patient in the supine position. A standard 5-port laparoscopic technique with the bed in the reverse Trendelenburg position is used. Patients not suitable for laparoscopy will undergo a traditional laparotomy (see Multimedia Appendix 1).

Table 1. TOTS variables and their assessment.

Items[a]	Baseline	Discharge	1 month	3 months	6 months	12 months	Annually
Demographic & medical history [b]							
Date of birth, sex, income, insurance	✓						
Marriage & education	✓						
Past medical history	✓						
Family history	✓						
Medications	✓	✓	✓	✓	✓	✓	✓
Behavioral & psychosocial assessment							
Diet and nutrition behavior	✓					✓	✓
Physical activity	✓					✓	✓
Quality of life	✓					✓	✓
Depression	✓					✓	✓
Smoking	✓						
Alcohol consumption	✓						
Drug abuse	✓						
Anthropometrics & physical examination							
Anthropometrics (height, weight, BMI, WC, HC, NC, wrist C)	✓		✓	✓	✓	✓	✓
Body composition (FM, FFM, LM)	✓		✓	✓	✓	✓	✓
General physical examination	✓		✓	✓	✓	✓	✓
Obesity-related comorbidities	✓		✓	✓	✓	✓	✓
Medical condition							
Cardiovascular (ECG, echocardiography)	✓					✓[c]	✓[c]
Respiratory (Chest X-ray, ABG, PFT, polysomnography)	✓					✓[c]	✓[c]
Gastrointestinal (endoscopy/barium meal, liver & gallbladder ultrasound)	✓				✓	✓[c]	✓[c]

Items[a]	Baseline	Discharge	1 month	3 months	6 months	12 months	Annually
Blood & urine assessments							
General blood biochemistry (CBC, LFT, LP)	✓				✓	✓	✓
Glucose homeostasis (FPG, HbA1C)	✓				✓	✓	✓
Hormonal assessment (TFT, PTH, Insulin)	✓					✓	✓
Micronutrients (Ca, P, Fe, Cu, Zn)	✓					✓	✓
Vitamins (D, B12)	✓					✓	✓
Inflammatory markers (CRP)	✓					✓	✓
24-hr urine albumin & creatinine	✓					✓[c]	✓[c]
Surgery							
General information (center, date, time, anesthetics)		✓					
Surgical procedure details		✓					
Additional/unprecedented procedures		✓					
Outcomes							
30-day surgical complications		✓	✓				
Long-term complications					✓	✓	✓
Re-admission[d]					✓	✓	✓
Reoperation					✓	✓	✓
EWL%			✓	✓	✓	✓	✓
Metabolic assessment			✓	✓	✓	✓	✓

[a]WC: waist circumference; HC: hip circumference; NC: neck circumference; Wrist C: wrist circumference; FM: fat mass; FFM: fat-free mass; LM: lean mass; ECG: electrocardiography; ABG: arterial blood gas; PFT: pulmonary function test, CBC: complete blood count:; LFT: liver function test; LP: lipid profile, FBG: fasting plasma glucose; HbA1C: hemoglobin A1C; TFT: thyroid function test; PTH: parathyroid hormone; CRP: C-reactive protein.

[b]Multimedia Appendix 1 for more information about the online forms.

[c]If necessary.

[d]Including in-patient and out-patient care.

Data Collection & Quality Control Procedures

An electronic database for precise data collection was designed. Manuals of operations and procedures were also created to minimize technical variability. Data collectors including study investigators and surgeons underwent training and certification with respect to study protocols. Quality control procedures including frequent contacts and visits between the surgeon and clinical center staff help to ensure complete and accurate data collection. Investigators used validated and standardized instruments for objective and subjective measures. When not available, new instruments were created to meet the specific goals of the study (see Multimedia Appendix 1).

A brief summary of each outcome domain and standard forms and measures used to assess each of these domains, as well as the contact points at which they will be administered, are described below.

Clinical Endpoints

In order to evaluate the effectiveness of bariatric surgery on obesity and obesity-related comorbidities, all participants are interviewed by a trained physician to complete a standardized clinical history questionnaire. It covers risk factors for cardiovascular disease, hypertension, hyperlipidemia, diabetes, and familial history of non-communicable diseases, smoking habits, drug abuse, and alcohol consumption. A similar postoperative survey is completed as well. The physicians are required to undergo periodic evaluation according to written protocols and control procedures to ensure up-to-date and universal practice.

Physical Examination and Anthropometric Measurements

Physical examination aims to look for obesity-related conditions as well as general health status of the individual. Anthropometrics include weight, height, neck, waist, wrist, and hip circumference, measured according to WHO guidelines [29]. Body composition is assessed by a portable bioelectrical impedance analyzer and output data includes body weight (kg), impedance (ohms), fat mass (kg), fat-free mass (kg), total body water (kg), and percent body fat (%) (see Multimedia Appendix 1).

Behavioral and Psychosocial Factors

The TOTS investigators have hypothesized that pre-existing psychological and behavioral factors could influence the outcomes after bariatric surgery. These aspects, such as quality of life and depression, will be assessed at baseline and follow-up, and will include questions on preoperative weight loss practices and eating habits (including binge eating and eating beyond satiation), tobacco use (according to US Centers for Disease Control and Prevention) [30], alcohol use, history of psychiatric disorders, and counselor/therapist contact. The depressive symptoms will be assessed using the Persian-language version of Beck Depression Inventory, version 1 [31,32]. Moreover, quality of life will be assessed by the Iranian version of Short Form Health Survey (SF-36) that measures eight health-related concepts, including the physical, mental, and social aspects of health [33]. Physical activity levels are assessed using the Persian-translated long form of International physical activity questionnaire (Persian IPAQ) [34]. The questionnaire measures all three forms of activities including leisure time, job, and household activities in the past week (see Multimedia Appendix 1).

Dietary Assessment

Diet plays a central role in the pathophysiology of obesity, as well as obesity treatment. Maintenance of weight loss after bariatric procedures is mainly achieved through changing dietary habits. In order to assess the role of diet on obesity before and after surgical interventions, an expert nutritionist assesses dietary intake of the patients using three consecutive 24-hour recalls, on weekdays (we selected weekdays because weekends do not reflect the usual diet of a patient). Portion sizes of meals are converted to grams by using household measures [35]. Nutrient intakes are calculated according to the US Department of Agriculture and Iranian Food Composition Tables [36,37].

Blood and Urine Biochemical Assessment

Blood and urine samples are collected before and after the surgery at 6 and 12 months, and annually thereafter. After 12-14 hours overnight fast, multiple aliquots of blood are drawn for biochemical and future genetic/molecular assessments. A standard 24-h urine collection is advised for all participants (see Multimedia Appendix 1).

Genetics and Biomarkers

DNA obtained from consenting participants will be part of an ongoing research effort by our team to identify genes related to human obesity. Blood samples drawn from subjects before and after the surgery are stored for further studies (see Multimedia Appendix 1).

Health Care Utilization

Studies addressing cost-effectiveness of bariatric surgery have shown controversial results and have not yet reached a universal conclusion [38-41]. In order to assess the financial burden of obesity in our setting, a dataset is designed to measure and evaluate short- and long-term cost-effectiveness of bariatric surgery pre and postoperatively.

Complications Related to Surgery

To document the frequency of complications after these most common techniques, as well as newer techniques in this study, a dataset is designed to describe early (occurring within 30 days of surgery) and long-term complications and factors associated with those events. Early complications known to complicate abdominal surgery include gastrointestinal adverse events (eg, anastomotic leak), thromboembolism, sepsis, and acute kidney injury. Long-term complications include gastrointestinal complications, cardiovascular events, and surgical re-intervention [42].

Follow-Up

All participants are scheduled for follow-up visits by a multidisciplinary team (ie, surgeon, obesity specialist, endocrinologist, and dietitian) at 10 days, and at 1, 3, 6, and 12 months, and annually thereafter. A trained nurse will ask about related medical conditions by means of email or over the phone (including cardiovascular, metabolic, pulmonary, renal,

musculoskeletal, urologic, reproductive, and gastrointestinal outcomes), and if a related event is noticed, the research investigator will actively seek respective data. In the case of mortality, data will be collected based on the death certificate.

Approval

This study has been approved by the Human Research Review Committee of the Endocrine Research Center, Shahid Beheshti University of Medical Sciences, No. 2ECRIES 93/03/13.

Results

This study has recruited 1050 participants as of September 2015 and is ongoing. Mean age of the participants is 37.8 years, mean BMI of 43.7 kg/m^2, with 76.9% female. Detailed characteristics of the participants including their baseline anthropometrics and prevalence of comorbidities are presented in Table 2.

Table 2. Baseline characteristics, anthropometrics, and laboratory values of the participants upon enrollment[a].

Variables[b]	Total (N=1050)	Female (N=807)	Male (N=243)	*P* value
Age, year, mean (SD)	37.8 (11.7)	38.6 (11.8)	35.2 (11)	.036
Age group, n (%)				<.001
<20	52 (4.2)	39 (4.1)	13 (4.9)	
20-29	229 (21.8)	162 (19.7)	67 (28.2)	
30-39	339 (32.8)	251 (31.8)	88 (36.7)	
40-49	241 (22.8)	197 (24.3)	44 (17.5)	
50-59	148 (14.7)	123 (15.8)	25 (10.9)	
60-69	39 (3.5)	34 (4.1)	5 (1.3)	
≥70	2 (0.2)	1 (0.1)	1 (0.4)	
Marital status, n (%)				.014
Single/never married	252 (24.1)	169 (25.2)	83 (37.6)	
Married	668 (61.7)	526 (62.7)	142 (58.4)	
Divorced	62 (6.2)	52 (6.4)	10 (4.1)	
Widowed	46 (3.6)	46 (5.7)	0	
Education, n (%)				.024
No education	10 (1.0)	7 (0.9)	3 (1.2)	
Primary school	27 (13.8)	23 (15.0)	4 (1.8)	
College	531 (50.7)	430 (52.2)	101 (45.9)	
University	448 (34.5)	321 (32.1)	127 (43.1)	
Employment status, n (%)				<.001
Unemployed	567 (54)	521 (64.6)	46 (18.9)	
Employed	458 (46)	261 (35.4)	197 (81.1)	
Smoking status, n (%)				<.001
Never smokers	819 (71.2)	661 (78.9)	158 (44.9)	
Current smokers	156 (15.4)	89 (11.2)	67 (30)	
Former smokers	143 (13.4)	80 (9.9)	63 (25.1)	
Hookah use, n (%)	163 (15.5)	91 (11.3)	72 (29.6)	.004
Alcohol consumption, n (%)	150 (14.3)	86 (10.7)	64 (26.3)	.004
Comorbidities, n (%)				
Hypothyroidism	186 (17.8)	169 (18.5)	17 (6.5)	<.001
Hypertension	174 (17.6)	137 (17.1)	37 (15.3)	
Dyslipidemia	168 (16.0)	137 (17.0)	31 (12.8)	
Arthritis	167 (15.9)	144 (17.8)	23 (9.5)	.039
Diabetes mellitus	158 (16.6)	127 (17.3)	31 (8.9)	
Cardiovascular disease	63 (6.0)	47 (5.8)	16 (6.6)	
Liver Enlargement	350 (92.1)	287 (72.4)	63 (83.5)	
Fatty liver	106 (27.5)	87 (27.4)	19 (27.5)	
Grade I	134 (35.2)	124 (41.4)	10 (17.1)	
Grade II	155 (40.7)	125 (41.6)	30 (45.4)	
Grade III	61 (16.2)	38 (17.0)	23 (33.3)	
Gallstones	34 (7.1)	30 (7.4)	4 (5.1)	

Variables[b]	Total (N=1050)	Female (N=807)	Male (N=243)	*P* value
Anthropometrics, mean (SD)				
Height, cm	163.5 (8.3)	161.2 (6.1)	175.5 (7.8)	<.001
Weight, kg (range 74-195)	118.6 (20.7)	114 (17.1)	142.6 (20.9)	<.001
BMI, kg/m^2 (range 32.4-79.3)	43.9 (7.1)	43.8 (5.9)	46.2 (5.5)	.09
BMI group (kg/m^2), n (%)				.054
25-29.9	9 (0.9)	8 (1)	1 (0.4)	
30-34.9	68 (6.3)	57 (7.4)	11 (4.4)	
35-39.9	243 (23.2)	197 (24.2)	46 (19.9)	
40-44.9	332 (32.1)	261 (32.5)	71 (30.7)	
45-49.9	235 (22.4)	176 (21.5)	59 (25.5)	
50-54.9	99 (9.3)	67 (8.2)	32 (13)	
55-59.9	42 (3.7)	24 (3.1)	18 (6.9)	
60-64.9	13 (1.3)	9 (1.2)	4 (1.7)	
65-69.9	7 (0.7)	6 (0.8)	1 (0.4)	
≥70	2 (0.2)	2 (0.3)	0 (0.0)	
Waist circumference, cm	125.1 (13.9)	122.5 (12.6)	138 (12.6)	<.001
Hip circumference, cm	133.6 (12.7)	133.5 (13)	134.4 (11.1)	.065
Neck circumference, cm	39.1 (3.9)	37.9 (2.8)	44.7 (3.4)	<.001
Wrist circumference, cm	17.9 (1.6)	17.6 (1.4)	19.5 (1.7)	<.001
SBP, mmHg	120.1 (16.7)	119.4 (16.6)	124.1 (16.5)	
DBP, mmHg	75.9 (11.8)	75.3 (11.6)	79 (12.7)	
Fat mass, %	49.8 (3.9)	50.6 (3.2)	46 (4.7)	<.001
Fat mass, kg	58.2 (11.9)	6.9 (10.8)	64.4 (15)	<.001
Fat free mass, kg	59 (11.4)	55.5 (7.5)	75.9 (12)	<.001
Laboratory values, mean (SD)				
FPG, mg/dl	107.9 (34.5)	109.4 (36.5)	99.6 (18.8)	<.001
HbA1C, %	5.6 (1)	5.6 (1.1	5.6 (0.7)	.037
TG, mg/dl	152.7 (76)	150.3 (70.9)	164.8 (97.7)	
Cholesterol, mg/dl	192.4 (38)	193.5 (37.4)	186.6 (41.4)	.033
HDL, mg/dl	49.6 (10.9)	50.2 (10.8)	46.3 (10.5)	.001
LDL, mg/dl	112.5 (32.2)	113 (31.7)	109.6 (35.0)	
Hemoglobin, g/dl	13.5 (1.5)	13.2 (1.2)	15.2 (1.4)	<.001
Hematocrit, %	41.0 (3.9)	40.2 (3.2)	45.7 (3.5)	<.001
AST, U/l	23.4 (15.5)	21.8 (12.8)	29.2 (18.6)	.007
ALT, U/l	28.7 (21.3)	26.1 (15.5)	41.3 (31.3)	<.001
Alkaline phosphatase, IU/l	191.8 (57.7)	191.4 (56.5)	194 (63.8)	

[a]For continuous variables, *t* test was used. For categorical variables, Pearson chi-square test was used.

[b]SBP: systolic blood pressure; DBP: diastolic blood pressure; FPG: fasting plasma glucose; HbA1C: hemoglobin A1C; TG: triglyceride; HDL: high-density lipoprotein, LDL: low-density lipoprotein; AST: aspartate aminotransferase; ALT: alanine aminotransferase

Discussion

The TOTS is a prospective longitudinal study evaluating many preoperative, operative, and post-operative aspects of bariatric surgery. High fidelity results are assured through the use of standard validated instruments. Results of this study will provide a comprehensive understanding of this growing medical condition and its treatment and will empower clinicians with evidence-based recommendations regarding patient selection and evaluation, surgery options, and follow-up care.

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Conflicts of Interest

None declared.

Multimedia Appendix 1

Supplemental information.

[PDF File (Adobe PDF File), 282KB - resprot_v5i1e8_app1.pdf]

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Abbreviations

BMI: Body Mass Index
BPD/BPD-DS: biliopancreatic diversion with or without duodenal switch
EWL: excess weight loss
LAGB: laparoscopic adjustable gastric banding
LSG: laparoscopic sleeve gastrectomy
RYGB: Roux-en-Y gastric bypass
TOTS: Tehran Obesity Treatment Study

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Protocol

A Multicenter Prospective Study to Investigate the Diagnostic Accuracy of the SeHCAT Test in Measuring Bile Acid Malabsorption: Research Protocol

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Abstract

Background: Bile acid malabsorption (BAM) is one possible explanation for chronic diarrhea. BAM may be idiopathic, or result from ileal resection or inflammation including Crohn's disease, or may be secondary to other conditions, including cholecystectomy, peptic ulcer surgery, and chronic pancreatitis. No "gold standard" exists for clinical diagnosis of BAM, but response to treatment with a bile acid sequestrant (BAS) is often accepted as confirmation. The SeHCAT (tauroselcholic [selenium-75] acid) test uses a radiolabeled synthetic bile acid and provides a diagnostic test for BAM, but its performance against "trial of treatment" is unknown. Fibroblast growth factor 19 (FGF-19) and 7-alpha-hydroxy-4-cholesten-3-one (C4) also offer potential new biomarkers of BAM.

Objective: This protocol describes a multicenter prospective study to evaluate the diagnostic accuracy of SeHCAT and 2 biomarkers in predicting BAM as assessed by trial of treatment.

Methods: Participating gastroenterology centers should have a minimum workload of 30 SeHCAT patients per annum. Patients should not be pregnant, on medication that could confound follow-up, or have any severe comorbidity. All eligible patients attending a gastrointestinal appointment will be invited to participate. On attending the SeHCAT test, blood and fecal samples will be collected for analysis of FGF-19 by enzyme-linked immunosorbent assay and for C4 and fractionated bile acids by liquid chromatography–mass spectrometry. A capsule containing radiolabeled SeHCAT will be administered orally and a scan performed to measure SeHCAT activity. Patients will return on day 7 to undergo a second scan to measure percentage SeHCAT retention. The test result will be concealed from clinicians and patients. BAS will be dispensed to all patients, with a follow-up gastroenterologist appointment at 2 weeks for clinical assessment of treatment response and adherence. Patients responding positively will continue treatment for a further 2 weeks and all patients will have a final follow-up at 8 weeks. The diagnostic accuracy of the SeHCAT test and biomarkers will be analyzed at different thresholds using sensitivity, specificity, positive and

negative predictive value, likelihood ratios, and area under the curve in a sample of 600 patients. Multivariable logistic regression models will be used to assess the association between presence of BAM and continuous SeHCAT retention levels after adjustment for confounders.

Results: Funding is being sought to conduct this research.

Conclusions: The SeHCAT test for diagnosis of BAM has been in common use in the United Kingdom for more than 30 years and an evidence-based assessment of its accuracy is overdue. The proposed study has some challenges. Some forms of BAS treatment are unpleasant due to the texture and taste of the resin powder, which may negatively affect recruitment and treatment adherence. Trial of treatment is not as "golden" a standard as would be ideal, and itself warrants further study.

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KEYWORDS

diagnostic test; accuracy; bile acid malabsorption; diarrhea; SeHCAT test; bile acid sequestrant

Introduction

Chronic diarrhea is a common problem and the investigation and management of the condition places a significant burden on health services as well as on affected patients. The suggestion that bile acid malabsorption (BAM) can cause diarrhea was first described by Hofmann in 1967 [1]; since then, BAM has been identified as a possible explanation for persistent chronic diarrhea.

Bile acids are produced in the liver, stored in the gallbladder, and released upon eating for the digestion of dietary fat. They are then largely reabsorbed by the terminal ileum and returned to the liver, a process known as enterohepatic circulation. When the reabsorption process is disrupted, excess levels of bile acids enter the colon where they lead to increased motility and water secretion, resulting in diarrhea.

Evidence is accumulating that BAM is more common than was previously thought [2,3]; however, robust data on the prevalence of BAM do not exist. Three types of BAM have been defined. In BAM Type 1, the bile acid malabsorption results from ileal resection, ileal disease including Crohn's disease, or bypass of the terminal ileum. BAM Type 2 relates to primary, idiopathic malabsorption, whereas BAM Type 3 represents malabsorption secondary to other conditions, including cholecystectomy, peptic ulcer surgery, chronic pancreatitis, and celiac disease.

A diagnostic test for BAM is provided by SeHCAT (tauroselcholic [selenium-75] acid), a radiolabeled synthetic bile acid. The SeHCAT test is a measure of the retention of radioactivity in the patient following administration of a capsule containing SeHCAT. The patient is scanned with a gamma camera 1 to 3 hours after taking the capsule and the scanning is repeated after 7 days to measure the percentage retention of the radiolabeled bile acid. A low SeHCAT retention level at day 7 represents an abnormal result for the test, indicating a positive diagnosis of BAM.

The SeHCAT test was introduced in the late 1970s; however, despite being used for more than 30 years, much of the available evidence is anecdotal knowledge built up over time rather than through systematic research. A recent survey on the use of SeHCAT in the United Kingdom provided an insight into the frequency of use of the test and the practicalities of its implementation in UK hospitals [4]. The study identified 73 centers using SeHCAT with a wide variation in the annual patient workload ranging from 1 to 300 tests (mean 51; median 30). An increase in referrals since 2010 was reported in response to demand from clinicians. Considerable variability in practical implementation of the technique was found, alongside a wide variation in the "normal" range of the SeHCAT percentage retention levels used for reporting, diagnosis, and treatment. The different approaches to definition of an abnormal result included a single threshold value (of which <15% retention was the most common), division into 3 categories (normal, borderline, and abnormal), or into 4 categories (normal, mild, moderate, and severe).

Patients with a diagnosis of BAM from a positive SeHCAT test may be offered treatment with bile acid sequestrants (BAS), such as cholestyramine, colestipol, and colesevelam. In general, patients who adhere to BAS treatment respond well and rapidly (within days) with significant reduction in bowel frequency and improvements in their quality of life [3]. However, certain BAS treatments are unpleasant to the patient and clear communication between the clinician and the patient is needed to highlight the expected benefits of treatment to ensure good adherence. Studies have found a relationship between SeHCAT retention levels and response to BAS treatment, with lower retention levels associated with greater resolution of symptoms [5].

A prospective survey was conducted in 2014 by the King's Technology Evaluation Centre (KiTEC) to characterize the clinical indications for referring patients for a SeHCAT test across 38 UK centers and to describe the range of test results and treatment pathways [6]. Patients with BAM Type 1 represented 14% of more than 700 patients tested with the remainder split fairly equally between BAM Types 2 and 3. Using center-defined thresholds, 51% of results were defined as abnormal or borderline; however, only 37% of patients were prescribed treatment with BAS. Median SeHCAT retention levels were much lower for BAM Type 1 (2%) compared with BAM Types 2 and 3 (18% and 17%, respectively).

The diagnostic performance of the SeHCAT test across the range of thresholds in current use is poorly understood. There is no established "gold standard" for the diagnosis of BAM. However, diagnosis is sometimes made through a trial of treatment with BAS and response to treatment with BAS offers a potential reference standard, if taken by *all* patients, not just those who the SeHCAT test suggests may be in the abnormal

range. A review by Riemsma et al [7] identified 3 studies which had taken this approach; however, the numbers were small (ranging from 13 to 46 patients) resulting in wide confidence intervals [7-10]. Specificity was more than 0.9 in all 3 studies; in the 2 studies that used a cut-off of 8% retention, sensitivity ranged from 0.67 to 0.95. Further research using larger samples is needed to assess the accuracy of the SeHCAT test in diagnosing the BAM condition as determined by response to BAS treatment.

In addition, 2 biomarkers have shown promising results as possible predictors of BAM, namely fibroblast growth factor 19 (FGF-19) and 7-alpha-hydroxy-4-cholesten-3-one (C4) [11,12]. Exploration of the performance of these biomarkers as alternative tools for discriminating between diagnoses could potentially improve diagnostic accuracy.

This protocol describes a study that will assess the diagnostic performance of the SeHCAT test and of 2 biomarkers in the prediction of BAM using a positive response to treatment with BAS as a definitive indication of the BAM condition. Improved diagnostic accuracy for BAM should, in turn, allow treatment of this debilitating condition to be optimized.

Objectives

The primary research objective is to investigate the diagnostic accuracy of the SeHCAT test in providing a positive indication of BAM among people with chronic diarrhea who are clinically suspected of having BAM, using trial of treatment with BAS as the gold standard.

The secondary research objectives are to investigate:

1. The continuum of results of the SeHCAT test, overall and for different clinical populations;
2. The feasibility of using the SeHCAT test to provide an indication of the severity of BAM (eg, mild, moderate, or severe);
3. Adherence to treatment with BAS, overall and for different clinical populations;
4. The diagnostic accuracy of 2 biomarker tests (FGF-19 and C4) in predicting BAM; and
5. Whether the biomarkers can provide information on the nature of BAM.

The tertiary research objective is, for established cut-off values, to compare the sensitivity and specificity of the SeHCAT test with those of the 2 biomarkers in providing a positive indication of BAM using trial of treatment with BAS as the gold standard.

The research objectives can be translated into the following research questions:

1. Is SeHCAT an accurate test to provide a positive indication of BAM and what are the optimal cut-off thresholds?
2. Can different cut-off thresholds be established for the different clinical populations under study?
3. How do the cut-off thresholds currently in use for SeHCAT in UK centers compare with these optimal values?
4. Is SeHCAT an accurate test to grade the severity of BAM?
5. Does adherence to treatment with BAS differ for the different clinical populations studied?
6. Can biomarkers provide an accurate test for a positive indication of BAM?
7. Is the accuracy of biomarkers higher than that of SeHCAT in giving a positive indication of BAM?

Methods

Type of Study

The protocol describes a multicenter prospective study. Patients referred for suspected BAM will undergo a SeHCAT test and be tested for the biomarkers before undergoing treatment with BAS. Response to BAS treatment will provide a gold standard indication of a diagnosis of BAM. The diagnostic accuracy of the SeHCAT test and of the biomarkers will be assessed for the prediction of BAM.

Setting

For a UK study, participating centers may be identified from the pool of 38 centers that took part in the recent KiTEC survey.

The proposed selection criteria for the centers are (1) a minimum workload of 30 SeHCAT patients per annum for the most recent calendar year for which data are available; (2) agreement to adopt a standardized SeHCAT test procedure; (3) laboratory capacity to collect, prepare, and deliver biological samples (blood and feces) for analysis at a remote site; and (4) formal commitment of a consultant gastroenterologist to participate in the study.

Study Population

Participants in the study will be adults recruited from a population of patients attending a secondary care gastroenterology appointment, presenting with chronic diarrhea of unknown cause, in whom BAM is considered clinically possible. Patients will be included if they are suspected of having BAM Type 1 (following ileal resection, ileal disease including Crohn's disease, or bypass of the terminal ileum), BAM Type 2 (primary idiopathic malabsorption), or BAM Type 3 (secondary to other conditions, including cholecystectomy, peptic ulcer surgery, chronic pancreatitis, and celiac disease). Potential participants will be screened for eligibility using a structured questionnaire and detailed clinical assessment.

Exclusion Criteria

Potential participants may not enter the study if ANY of the following apply:

1. They are unable to provide informed consent;
2. They are on medication that could confound follow-up assessment;
3. They are pregnant or they are at risk of pregnancy and do not wish to take adequate precautions against pregnancy for the duration of the study;
4. They have any severe comorbidity condition with less than 12 months life expectancy;
5. They are unable or unwilling to undergo a SeHCAT procedure following the standard protocol and/or to provide blood and fecal samples; or
6. They are unwilling to undergo a course of treatment with BAS.

Study Procedure and Outcome Measures

A diagram illustrating the study procedure is shown in Figure 1. All patients attending a gastrointestinal appointment at a participating center and fulfilling the inclusion criteria will be invited to participate in the study. Individual patient consent will be requested at entry by the recruiting clinician. All patients who fulfill the inclusion criteria will be given detailed information both verbally and in the form of a patient information sheet. During the gastrointestinal appointment, patients will be referred by the clinicians for a SeHCAT test. The patients will be given detailed information of all relevant pretest and posttest dietary and medication requirements.

When the patient visits the hospital for the SeHCAT test, blood and fecal samples will be collected before the test using standard clinical procedures. The samples will be transferred to the local biochemistry laboratory at each participating site, where they will be handled by a trained biomedical scientist and prepared for transfer to the reference laboratory. The preparation procedure will involve taking a blood sample in a 5-mL tube containing no anticoagulants. This sample will be centrifuged in a standard bench-top centrifuge for 15 minutes at 3000 rpm, and the resulting serum separated from the cells and stored at –20°C before shipment to the central laboratory. A random fecal sample will be collected and stored at –20°C before shipment. The samples will be shipped on dry ice to the reference laboratory, where the serum samples will be analyzed for FGF-19 by enzyme-linked immunosorbent assay (ELISA), and for C4 and fractionated bile acids by liquid chromatography–mass spectrometry (LC-MS) using established techniques.

The SeHCAT test will be performed in the Nuclear Medicine Department of each participating center, following a standard protocol, under the supervision of an Administration of Radioactive Substances Advisory Committee (ARSAC) certificate holder [13]. A capsule containing radiolabeled SeHCAT will be administered to the patient orally according to a standard procedure and considering manufacturer's recommendations. Patients will receive specific instructions regarding their food intake. After 3 hours, a scan will be performed to measure the baseline SeHCAT activity.

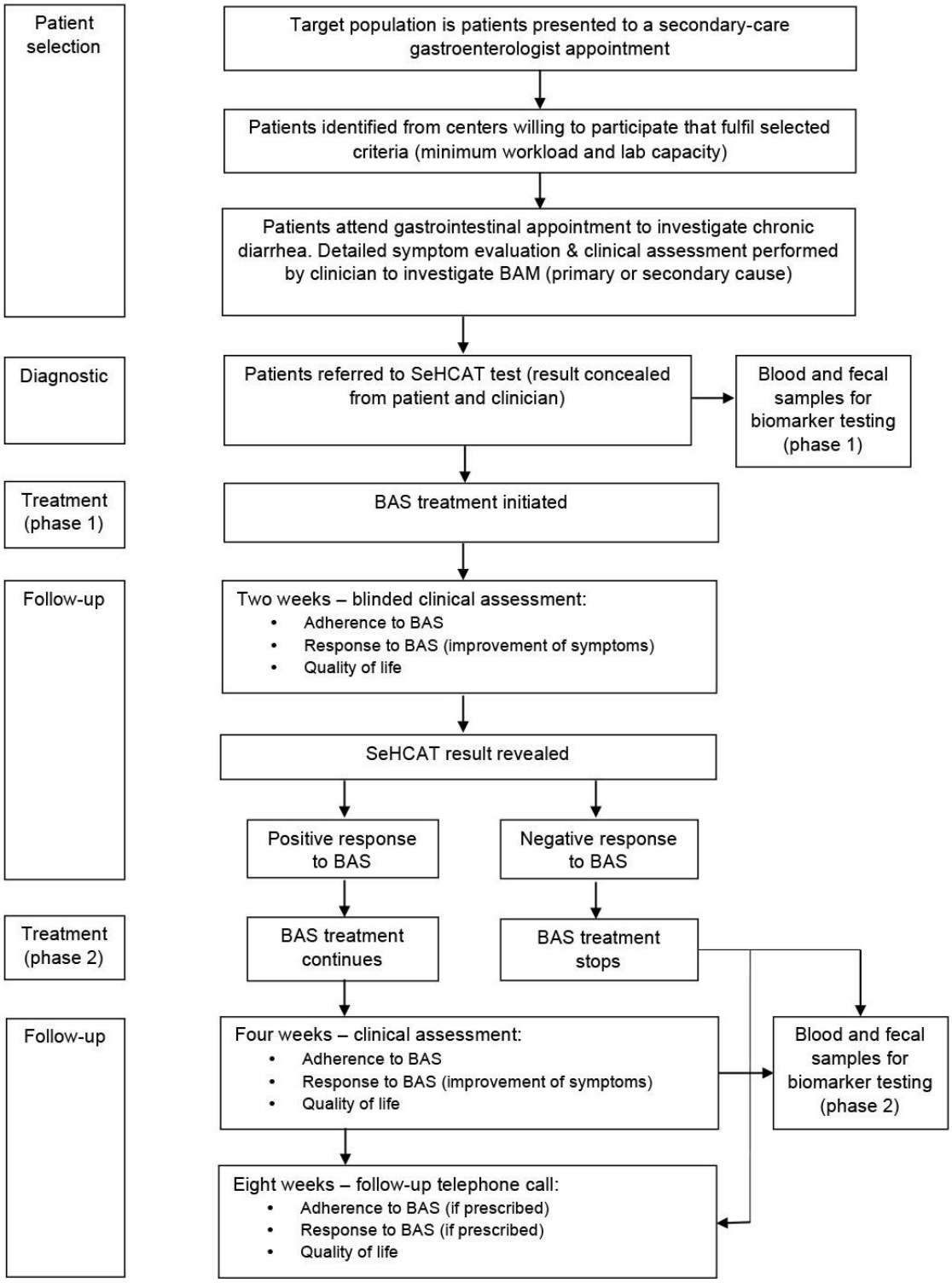
The patient will be requested to return to the Nuclear Medicine Department on day 7 to undergo a second scan to measure the retention of SeHCAT, thus completing the test. The SeHCAT test result will be concealed from the clinician and the patient until the 2-week follow-up appointment. On the visit to complete the SeHCAT test, BAS will be dispensed to allow the patient to commence treatment on the following day. BAS will be prescribed to all patients according to a standard protocol based on current clinical practice.

All patients will attend a follow-up appointment with the gastroenterologist 2 weeks after the initiation of treatment with BAS. A clinical assessment will be performed to capture the relevant outcome measures: the patient's adherence to treatment, improvement of symptoms, and quality of life (QoL). The QoL assessment will be undertaken by a research nurse trained in qualitative methodology. The clinical assessment will determine whether there has been a positive or negative response to BAS treatment. A standardized definition will be agreed for the degree of improvement in symptoms required to represent a positive response to treatment. At the follow-up appointment, the SeHCAT test result will be revealed to the clinician and the patient. A positive response to treatment at 2 weeks with evidence of treatment adherence will be used as the gold standard that defines a diagnosis of BAM.

Patients with a negative response to treatment will stop BAS treatment promptly (irrespective of SeHCAT test result) and may be directed to other investigations. At this stage, blood and fecal samples will be collected from this group for assessment of biomarkers. Patients with a positive response to BAS treatment (irrespective of SeHCAT test result) will continue on BAS treatment for a further 2 weeks. For this group, a second follow-up appointment will be performed at 4 weeks to repeat the outcome assessment. At this second follow-up visit, blood and fecal samples will be collected for further biomarker testing.

At 8 weeks, all patients entered into the study will be contacted for a telephone follow-up appointment to collect final follow-up data. Any interventions other than those included in the study protocol will be recorded during the study period.

Figure 1. Study procedure.



Data Collection

Relevant clinical data regarding the care pathway, diagnostic tests, and interventions will be collected using electronic case report forms. Patient follow-up data will be collected using a questionnaire to be developed according to the Diagnostic Criteria for Functional Gastrointestinal Disorders ("the Rome criteria") [14]. Quality of life measures will be captured using the standard EQ-5D tool [15].

All study data will be entered onto a database by qualified research staff and subjected to quality control checks. Patient-identifying details will not be included in any study data electronic files. After the closure of the study, the participating sites will maintain all source documents, study-related documents, and copies of the paper source documentation forms, data query, and amendment forms in compliance with local center policies. All source documents will be retained for a period of 10 years following the end of the study.

Sample Size

Sample size was calculated based on the 95% confidence intervals of the sensitivity and specificity of the SeHCAT test result in predicting true BAM as defined by response to BAS treatment [16]. The prevalence of BAM in this population of patients attending gastroenterology appointments with chronic diarrhea is estimated to be approximately 50% [6]. Assuming a minimum value of 70% for either sensitivity or specificity [7], a sample of 600 patients (and therefore 300 true positives) would give an acceptable 95% confidence interval of 65% to 75%. Sensitivities or specificities closer to 1 will have narrower confidence intervals. If the assumption of 50% true positives is incorrect, an alternative scenario of 3:1 positives to negatives would give a 95% confidence interval of 62% to 77% around a specificity of 70%.

Statistical Analysis

Results of the SeHCAT test (percentage retention per patient) will be presented using suitable descriptive methods, including frequencies, proportions, means, and standard deviations, as appropriate. Response to BAS treatment will be presented as a proportion of those who initiated the treatment. All descriptive statistics will be presented with 95% confidence intervals.

The diagnostic accuracy of the SeHCAT test to provide a positive indication of BAM compared with the gold standard of a positive response to BAS at 2 weeks of treatment adherence will be investigated using the receiver operating characteristic (ROC) curves approach to compare different choice of thresholds. Sensitivity, specificity, positive predictive value, negative predictive value, likelihood ratios, and area under the curve will be calculated and presented with 95% confidence intervals. Multivariable logistic regression models will be used to assess the association between presence of BAM and continuous SeHCAT retention levels after adjustment for confounders.

Similar methods will be used to assess the diagnostic accuracy of biomarkers in the prediction of BAM. The diagnostic accuracy of the selected biomarkers will be compared to that of the SeHCAT test using the McNemar test for paired proportions.

Where diagnosis of BAM is confirmed, the association between severity of BAM (as assessed clinically at baseline) and the continuous SeHCAT result will be investigated using appropriate multivariable regression models taking into account prognostic baseline variables as potential confounders.

The EQ-5D scores [15] will be derived using standard procedures and will be used to describe the QoL of different subgroups. The change in QoL for the SeHCAT-positive group who respond positively or negatively to BAS will be examined and compared using regression methods adjusting for prognostic variables including QoL at baseline.

All analyses will be 2-sided with 5% significance level and will be reported according to the international standards for the reporting of diagnostic accuracy studies [17].

Ethics and Governance

Individual patient consent will be requested at entry by the recruiting clinician. All patients who fulfill the inclusion criteria will be given detailed information by a qualified researcher both verbally and in the form of a patient information sheet. Patients will be able to withdraw consent at any time during the study. Patients may also be withdrawn from the study for medical reasons.

Permission and approval for the proposed research will be requested using the relevant procedures for ethical review of studies of NHS patients in the United Kingdom. The protocol will be reviewed by an NHS Research Ethics Committee, overseen by the UK Health Research Authority. Local research and development approval will also be obtained from each participating center.

The chief investigator will be responsible for supervising the conduct of the research and for protecting the rights, safety, and welfare of the participants enrolled in the study. Principal investigators will be identified at each participating site to ensure that the research activities are conducted in an ethical manner and in accordance with UK regulations, institutional policies, and good clinical and research practice.

Safety and Adverse Incidents

All adverse events will be recorded and acted on by a qualified member of staff (eg, a nurse under the supervision of the clinician). The SeHCAT test is standard practice and is a safe test that has been in use in the United Kingdom for more than 30 years. The SeHCAT radiolabeled synthetic bile acid is licensed and approved for use in the United Kingdom. The test involves exposure to a low dose of ionizing radiation. All procedures will comply with ARSAC regulations and patients will be given radiation protection instructions both verbally and in writing. The BAS drugs to be prescribed are licensed and used in standard practice, and will be used in full compliance with the licensed indication.

Study Committees

A multidisciplinary Study Management Group will be responsible for running the study on a day-to-day basis and will include a manager, chief investigator, clinicians, and statisticians. A Study Steering Committee will take major decisions, such as changing the protocol, and will include members who are not involved with the running of the study. A Data and Safety Monitoring Committee will be established and will consist of at least 3 independent members, one of whom will be a clinical specialist and one a statistician. This committee will be responsible for assessing recruitment data and study conduct and the monitoring of adverse events. The research team will include external independent members, including a patient representative on the committees, to aid the team in making unbiased decisions free from financial, personal, or professional pressure.

Results

Funding is currently being sought to carry out this proposed research.

Discussion

Strengths

The SeHCAT test for diagnosis of BAM has been in common use in UK nuclear medicine centers for more than 30 years. An evidence-based assessment of the accuracy of the SeHCAT test is overdue. This research is seen as a priority by the National Institute for Health and Care Excellence in the United Kingdom [18], who funded the recent prospective study of UK centers [6] and the development of this protocol.

Chronic diarrhea is a debilitating condition that is not only physically unpleasant to endure, but impacts on social engagement and quality of life. Improving diagnostic accuracy in the prediction of BAM should help lead to optimization of treatment for those patients whose diarrhea is linked to this condition and clinicians will be able to advise patients with greater confidence.

The recent UK review [4] and survey [6] suggest that the timing is currently optimal for conducting the proposed research. Up-to-date contextual baseline data are available and UK centers have recently engaged in the provision of data, with a database of contacts available for centers, all of which should improve the feasibility of conducting the study.

Challenges

A positive response to BAS treatment is considered here as the gold standard for the diagnosis of BAM; a negative response will be coded as the patient not having BAM. However, this assumes that BAS is always effective in patients suffering from BAM. Without further research, it is impossible to assess whether this is a realistic claim. Similarly, it is possible that patients may have a placebo response (ie, an improvement in clinical symptoms even if they do not have underlying BAM). Longer-term follow-up may help to assuage these concerns.

Some forms of BAS treatment are unpleasant and poorly tolerated, including cholestyramine and colestipol, due to the texture and taste of the resin powder. This may reduce both the willingness of patients to take part in the study and adherence to treatment once started. Recruitment may also be affected by the fact that *all* patients will receive BAS regardless of whether SeHCAT indicates any likely benefit. However, in this regard, maintaining blindness to the SeHCAT result should help to retain participants once they have agreed to take part. These issues could be surmounted by using the drug colesevelam, which is available in tablet form and is well tolerated. However, colesevelam has not been assessed in randomized controlled trials for the treatment of BAM and is not currently licensed in the United Kingdom for this purpose [19].

Conducting a prospective multicenter study presents its own challenges, which include potential variation in procedures and in data quality between centers and the need for effective management of centers to ensure study timelines are achieved. To address these challenges, standardized protocols will be developed for the administration of the SeHCAT test and the laboratory assessment of biomarkers, and definitions will be agreed for all clinical assessments, including those required to assess patient eligibility and response to treatment. A consistent approach will be developed for communicating to patients the potential benefits as well as adverse effects of BAS treatment to encourage optimum treatment adherence. The study manager will be responsible for liaising with centers to ensure that agreed procedures are observed, that patients are recruited and followed up, and that data are complete and uploaded to the database in a timely fashion.

Future Directions

The results from this study will need to be validated in other populations in the future. Populations with different dietary content, such as proportion of fiber, may react differently to the SeHCAT test [8]. Analysis of the variation in SeHCAT results between different centers will give an indication of the extent to which population-specific approaches may be required.

Depending on the results found for the 2 biomarkers considered, other biomarkers may be worth investigating as well as the potential for combining the SeHCAT and biomarker results.

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Authors' Contributions

JP and JS led the development of the design of the study protocol and drafting of the initial manuscript. FR led on writing the final manuscript. All authors participated in critical review of the methods and read and approved the final manuscript.

Conflicts of Interest

None declared.

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Abbreviations

C4: 7-alpha-hydroxy-4-cholesten-3-one
ARSAC: Administration of Radioactive Substances Advisory Committee
BAM: bile acid malabsorption
BAS: bile acid sequestrant
ELISA: enzyme-linked immunosorbent assay
FGF-19: fibroblast growth factor 19
KiTEC: King’s Technology Evaluation Centre

NIHR: National Institute for Health Research
QoL: quality of life
SeHCAT: tauroselcholic (selenium-75) acid

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Protocol

Same-Day Counseling: Study Protocol for the Evaluation of a New Mental Health Service

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Abstract

Background: Single-session counseling is being implemented across Canada to increase the accessibility and availability of mental health services. Despite increasing use, existing research on single-session counseling is sparse and has methodological limitations. In addition, some stakeholders are skeptical that this model of care can support meaningful change for clients.

Objective: The aim of this study is to evaluate a new single-session counseling program (called Same-Day Counseling) offered in an outpatient community mental health clinic in Northwestern Ontario, Canada.

Methods: Clients who attend Same-Day Counseling services will be given the opportunity to participate in the program evaluation. Those who consent will complete measures before their session, after their session, and at 1-month follow-up. Data will provide information on who accesses Same-Day Counseling (eg, typical presenting problems, symptom severity), client satisfaction with services, and whether clients benefit from the services (eg, improved functioning and reduced symptom severity).

Results: Data collection is underway with 80 participants having completed baseline measures and 55 participants having completed follow-up measures. Data collection is expected to conclude in December 2015.

Conclusions: This study is designed to contribute to the literature regarding the integration of single-session counseling into ongoing mental health services, with additional attention to methodological rigour. Our approach will help to address ongoing concerns regarding the implementation of single-session counseling, and inform health care providers and policy makers regarding the utility of this model for addressing the mental health care need of the community.

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KEYWORDS

single-session counseling; mental health services; outpatient; program evaluation; access

Introduction

Current State of Mental Health Services

Many Canadians are in need of mental health services, with these needs often going unmet, especially in the public system [1]. Service providers and administrators are acutely aware of the demand, yet struggle with limited resources available to provide the necessary care. As a result, mental health service agencies in Canada have long wait times (eg, up to 2 years) with some programs forced to temporarily shut their doors due to an inability to meet increasing demands [2-4]. This situation can result in deterioration of clients' mental health conditions and increased reliance on acute medical care services (eg, emergency departments) [5]. Novel approaches to meeting clients' needs are required to improve the accessibility and the availability of mental health services given the current resource limitations.

Once clients are registered with mental health services, high rates of missed appointments (eg, 15%) and dropouts (eg, 20%) are common and are related to reduced treatment outcomes [6,7]. For counselors, nonattendance initially results in underutilization of their time [6,7], as the missed session could have been made available to other clients. Afterwards, additional time is spent contacting clients who did not attend, which further restricts counselors' time to see clients [6]. Innovative service solutions that address nonattendance issues are necessary in order to increase the efficiency of mental health services.

Difficulty determining treatment sufficiency in counseling also contributes to service inaccessibility. It is often unclear to counselors whether termination of ongoing care is appropriate [8,9]. Research into effective psychotherapy duration traditionally suggested a linear relationship between number of sessions and treatment outcomes, where a greater amount of counseling results in greater improvements [10]. Recent research, however, demonstrates that while longer durations of counseling are more effective overall, rapid improvement occurs early in treatment, with each additional session producing less significant results [11,12]. In fact, if clients have not experienced functional changes by the eighth session, the likelihood of significant change is greatly diminished [11]. This more recent understanding of rates of improvement throughout the course of counseling further highlights the need for novel approaches to service provision.

Single-Session Counseling

One model of care, single-session counseling, has been implemented in an attempt to increase the availability and accessibility of mental health services. This type of counseling is broadly defined as any therapeutic encounter determined at the outset to be self-contained by both the counselor and the client [13]. The session is approached as a single encounter, regardless of the client's intention to access the service in the future. Single-session counseling has been offered by mental health professionals through community mental health centers and counseling centers in Canada, the United States, Europe, and Australia, and is also available in primary care and educational settings [8]. It is predominantly utilized in the form of walk-in counseling, however, some programs give clients the opportunity to schedule an appointment on the day they would like to attend [8]. With most single-session counseling programs, the option to return for future sessions is presented [8]. Clients who access single-session counseling multiple times may or may not meet with the same counselor, as many programs employ a team approach [3,4]. Screening for more severe presenting problems, such as suicidal or homicidal ideation, occurs frequently within this model of care with the hope that these clients will be directed to alternative crisis management services.

Single-session counseling is often associated with a solution-focused or client-centered approach to service delivery [14,15]. In this form of counseling, clients are in charge of the frequency in which they seek out support from a counselor, thereby being more client-driven and increasing sense of control for the client. As a result, the single-session counselor is permitted to be resourceful and flexible in using a variety of counseling modalities to best meet the needs of the clients in the moment [14,15]. While single-session counselors report having a greater sense of urgency to clarify priorities during each contact [15], they vary in their approaches to accomplishing this goal.

Different procedures are utilized to integrate single-session counseling into currently existing mental health services. One option is to replace the intake appointment with single-session counseling [4]. Single-session counseling can also be provided to clients that are on the waitlist for future services [16]. Other programs operate as stand-alone services with no direct connection to additional programming. Flexibility in the implementation of single-session counseling, and incorporation into ongoing mental health services, has resulted in an increase in this model of care in Ontario, Canada [3].

Integration of single-session counseling into current services has the potential to address many issues faced by mental health service providers. There is little to no wait for services with single-session counseling, allowing clients to access services when motivation and need are highest. Dropouts do not exist in single-session counseling, as there is no expectation of ongoing sessions. The self-contained format, along with the short-term nature of the scheduling process, also greatly reduces rates of missed appointments. Finally, treatment sufficiency is no longer an issue, as each session of counseling is considered sufficient, regardless of whether the client returns for future sessions [8,9]. Due to these factors, the single-session counseling format presents a possible option for meeting the current needs of clients, mental health care providers, and program administrators.

Available Evidence on Single-Session Counseling

Although research is limited, there is preliminary evidence for the utility of single-session counseling in reducing barriers to services, increasing client satisfaction, and helping clients address their mental health concerns. A review conducted by Hymmen et al [8], found anywhere from 74% to 100% of clients are satisfied across a wide range of single-session services. Also, 61% of clients found a single session of counseling sufficient to meet their needs and that they did not require additional sessions [8]. Single-session counseling is associated with symptom reduction and improvement in coping with presenting problems [8,9]. Receiving helpful advice about the problem, having the opportunity to talk about the problem and feel supported, and the immediate accessibility of mental health services were identified as helpful aspects of single-session counseling [8]. Findings are mixed regarding the relationship between type and severity of the presenting problem and outcomes in single-session counseling [8].

Despite these promising results, additional evidence is needed. Moreover, the available research has methodological issues that reduce the ability to draw firm conclusions about the utility of single-session counseling. For example, most services lack ongoing standardized outcome measurement for evaluation, relying mainly on client satisfaction questionnaires, and/or employee-created measures that lack psychometric data on their reliability and validity [8]. Another limitation is the involvement of the counselors in data collection and analysis, which may

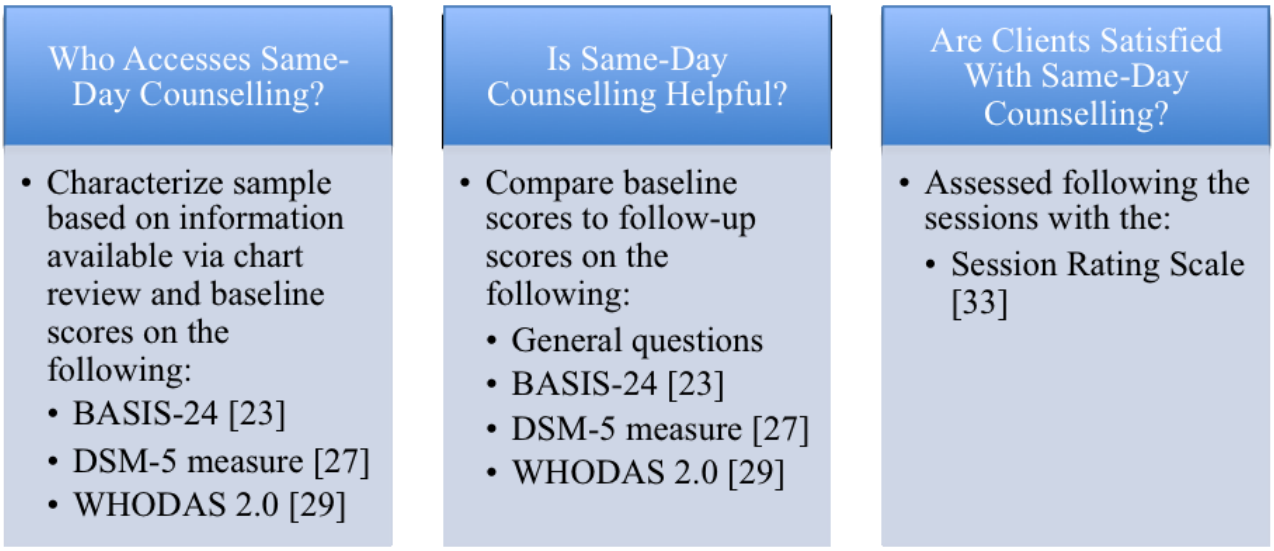
lead to response bias among participants [8]. Evaluations have also exhibited high attrition rates (eg, upwards of 50%) [17], which may be attributable to the method of data collection [3]. Lastly, restrictions on who can access services (eg, limited to clients without suicidal or homicidal ideation) reduce the generalizability of the results to a larger client base [3,8]. These limitations result in uncertainty regarding the utility of this service model and further emphasize the need for additional research.

In addition to the limitations of existing evidence, some counselors and decision makers have reservations about brief interventions [4,18], believing that they do not address the underlying problem [19]. Some counselors concede that single-session counseling can be effective, however, view it as ineffective or even ill-advised for clients with more complex or severe presenting problems [8,14]. Additional concerns exist that the increased presence of single-session counseling programs is due to demands placed on health service providers and is not in the best interest of the clients [19]. Further evidence is necessary to support the use of single-session counseling and validate its integration into mental health services.

Aims and Hypotheses

This paper describes the protocol our team will use to evaluate a new single-session counseling program (called Same-Day Counseling) offered in an outpatient community mental health clinic in Northwestern Ontario, Canada. Our study was designed with increased attention to methodological concerns including the use of multiple standardized measures, exclusion of the counselor from data collection and analysis, use of phone calls for follow-up evaluation to reduce attrition from the evaluation, and a lack of restrictions on who can access the program. Participants will be characterized in terms of demographic variables, symptomatology, contact with mental health services, and wait times. In addition, this evaluation will examine not only client satisfaction, but also changes in scores on measures of mental health functioning, psychiatric symptoms, and general health functioning. It is hypothesized that participants will be satisfied with the service, and their ability to manage their presenting problems as well as their mental health, general health and functioning will increase, and their symptoms will decrease. The evaluation questions, along with the corresponding measures and domains being assessed, are presented in Figure 1.

Figure 1. Summary of evaluation questions, domains assessed, and outcome measures included.



Methods

Evaluation of a New Program

Our study will evaluate a novel single-session counseling service, referred to as Same-Day Counseling, using a single group pre-post design. Same-Day Counseling adheres to the single-session service model, in that each session is considered self-contained. The Same-Day Counseling program is provided in a prescheduled format in which clients book an appointment for the day they would like to attend by calling reception staff that morning and selecting an available time that day (ie, they call in on the "same-day" they would like to attend). Clients are able to access the service as often as they would like, and have the option of meeting with the same counselor should they attend multiple sessions. There are no restrictions on who can access the service in terms of symptom severity or presenting problem.

Setting

The program evaluation will take place at a large outpatient community mental health clinic in Thunder Bay, Ontario, Canada (ie, St Joseph's Care Group Mental Health Outpatient Programs). Thunder Bay is a city in Northwestern Ontario that has a population of about 108,000 and operates as a hub for health care in the region [20,21]. Although rurally located, this city contains a regional hospital, an international airport, as well as a college and a university.

The clinic where the evaluation will take place serves adults with serious and persistent mental health problems and promotes the principles of psychosocial rehabilitation and recovery within an evidence-based and best practices model of care [22]. Services offered throughout the program include assessment, individual counseling, group counseling, medication management, spiritual care, referrals to community resources,

advocacy, and primary health care as required [22]. Self-referrals and referrals from other service providers (eg, family physicians, nurse practitioners) are accepted. Services are publically funded through the Ontario government, and provided at no cost to the clients upon presentation of an Ontario Health Insurance Plan card.

Participants

Clients registering for outpatient mental health services who are not yet connected to an individual counselor will be provided with information regarding the Same-Day Counseling program. Those who pursue Same-Day Counseling will be informed of the evaluation and invited to participate. There are no exclusion criteria. For participating in the evaluation, clients will be compensated with a US $10 gift certificate to a local coffee shop and entered into one of 5 draws for a US $100 grocery gift certificate.

Intervention

Same-Day Counseling sessions are typically 60-90 minutes in length and focus on addressing clients' immediate mental health concerns. Counselors are registered social workers with previous experience providing counseling within the clinic. Incoming counselors are given a general overview of the model and the rationale behind the service. No formal training was completed regarding the provision of Same-Day Counseling, as they are expected to use their clinical judgment and evidence-based interventions as they would with traditional forms of therapy. In general, interventions delivered include techniques from cognitive-behavioral therapy, dialectical behavior therapy, and emotion-focused therapies, among others. Counselors also assist clients with functional tasks associated with goal setting, securing safe housing, navigating legal matters, and promoting health and wellness. Counselors do not book follow-up appointments with clients, but welcome clients to attend Same-Day Counseling in the future, as needed.

Outcome Measures

The research team reviewed relevant literature and consulted with the counselors when choosing the outcome measures. Considerations included breadth and depth, length, clinical utility, accessibility, as well as their psychometric properties. Researchers generated 4 general questions (see Multimedia Appendix 1) that will be used to assess participants' ability to manage their presenting problems including the amount of stress the problem is causing, the amount of understanding participants have related to the cause of the problem, the amount of confidence participants have to cope with the problem, and the amount of knowledge, supports, or resources participants have to manage the problem.

The Behavior and Symptom Identification Scale-24 (BASIS-24) [23] will be included as a measure of mental health symptoms and functioning. This measure provides an overall score as well as subscale scores for the following domains: depression, interpersonal relationships, psychotic symptoms, alcohol/drug use, emotional lability, and self-harm [24]. The BASIS-24 displays excellent psychometric properties in outpatient samples (eg, internal consistency alpha =.70-.81; concurrent validity r=.59-.82) [25,26].

The Diagnostic and Statistical Manual-5 Level 1 Cross Cutting Symptom Measure for Adults (DSM-5 Measure) will be used to assess a variety of domains of psychiatric symptom frequency and intensity, including depression, anger, mania, anxiety, somatic symptoms, suicidal ideation, psychosis, sleep problems, memory, repetitive thoughts and behaviors, dissociation, personality functioning, and substance use [27]. The DSM Measure is a reliable instrument in North American populations (eg, test re-test reliability r=.66-.97) [28].

The World Health Organization Disability Assessment Schedule 2.0 (WHODAS 2.0) will be included as a measure of general health, functioning, and impairment [29]. The following six domains of functioning are assessed: cognition, mobility, self-care, getting along, life activities, and participation in society [30]. The WHODAS 2.0 has been used worldwide, and is psychometrically validated for use among individuals with common mental health disorders (eg, internal consistency alpha=.89; convergent validity r=.58) [31,32].

Finally, the Session Rating Scale (SRS) will be used as a measure of client satisfaction. The SRS is a brief global measure of therapeutic alliance designed to be easily administered in a clinical setting [33], while maintaining reliability and validity for individuals accessing outpatient mental health services (eg, internal consistency alpha=.88; convergent validity r=.48; test re-test reliability r=.64-.70) [34].

Procedure

Ethics approval for the study was obtained through a hospital research ethics board (ie, St Joseph's Care Group) and a university research ethics board (ie, Lakehead University). All participants will be informed that participation, nonparticipation, or withdrawal from the evaluation will not affect the care received. Documentation of participants' involvement in the study will not be included in their clinical chart, and counselors will not be involved in recruitment, data collection, or data analysis. To maintain confidentiality, personal identifying data will be stored separately from evaluation information. Only the researcher team will be able to access both files.

Clients attending Same-Day Counseling who consent to participate in the evaluation will be given a questionnaire package by reception staff. Measures completed pre-session, post-session, and at 1-month follow-up are presented in Table 1.

Table 1. Timeline for administration of outcome measures.

Presession	Postsession	One-month follow-up
General questions	General questions	General questions
	Session Rating Scale (SRS)	
Behavior and Symptom Identification Scale-24 (BASIS-24)		BASIS-24
World Health Organization Disability Assessment Schedule 2.0 (WHODAS 2.0)		WHODAS 2.0
Diagnostic and Statistical Manual-5 Self-Rated Level 1 Cross Cutting Symptom Measure for Adults (DSM 5 Measure)		DSM 5 Measure

Following their session, participants will leave the completed questionnaire package with reception staff. Approximately 1 month after their session, participants will be contacted by phone by a research assistant to complete the follow-up measures. Also after the session, the research team will conduct a chart review to characterize the individuals accessing Same-Day Counseling in terms of demographic and service utilization information.

If a participant endorses suicidality on the evaluation questionnaire, he or she will be informed by a research assistant that his or her counselor will be in touch to assess the current level of risk and to provide immediate support. Crisis response contact information will also be provided. A counselor will attempt to contact the participant by phone within 24 hours to further assess the risk. This protocol is put in place to keep clients safe and to ensure that researchers do not enter into dual relationships with the clients (eg, researcher and counselor).

Statistical Analyses

Descriptive statistics will be conducted for participants in the Same-Day Counseling program to quantify demographic variables, symptomatology, contact with mental health services, and wait times, as well as participants' ratings of the counseling session. Paired-samples *t* tests will be used to compare pre-session and follow-up scores on participants' ability to manage the presenting problem, their psychiatric symptom frequency and intensity, their mental health and functioning, and their general health and functioning.

Sample Size Calculation

A power analysis was conducted using G*Power Software to determine the required sample size. Given a 2-sided alpha level of .01, a power of .8, and an intended effect size of .56, the required sample size is 41 participants. The chosen effect size was informed by research on the psychometric properties of the BASIS-24 [24]. Rates of attrition in single-session counseling service evaluations vary greatly depending on the type of follow-up contact and the duration between baseline and follow-up [8]. Based on an expected attrition rate of 30%, the current evaluation requires a sample size of 59 participants [35]. Participants will be recruited from the Same-Day Counseling program until sufficient sample size at follow-up has been achieved.

Results

Participant recruitment began in February 2014. Currently, 80 participants consented and completed pre- and post-session measures; 18 of the participants completed the baseline questionnaires on multiple occasions, for a total of 110 completed baselines. So far, 55 participants have completed 1-month follow-up measures. Data collection is expected to conclude in December 2015, with an estimated 76 participants, based on the current rate of attrition.

Discussion

Single-session counseling is a potential option for meeting the needs of clients, one that has been available throughout Canada for a number of years. Program evaluations of single-session counseling services provide preliminary evidence supporting its use, but often lack attention to methodology. Additional research is required to ensure that the continued implementation of this model of care is substantiated by evidence.

The results of this study will contribute to the research on integration of single-session counseling into ongoing mental health services, and hopefully encourage further research into such programs. This evaluation will also attempt to address the ongoing concerns that implementation of single-session counseling may be motivated by factors other than the best interest of clients. Ultimately, additional literature on single-session counseling would help inform service providers and policy makers surrounding the best use of mental health service resources.

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Conflicts of Interest

None declared.

Multimedia Appendix 1

General Questions.

[PDF File (Adobe PDF File), 267KB - resprot_v5i1e22_app1.pdf]

Multimedia Appendix 2

Funding approval.

[PDF File (Adobe PDF File), 137KB - resprot_v5i1e22_app2.pdf]

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Abbreviations

BASIS-24: Behavior and Symptom Identification Scale-24
DSM 5 Measure: Diagnostic and Statistical Manual-5 Level 1 Cross Cutting Symptom Measure for Adults
SRS: Session Rating Scale
WHODAS 2.0: World Health Organization Disability Assessment Schedule 2.0

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Protocol

Multiple Sclerosis Therapy With Disease-Modifying Treatments in Germany: The PEARL (ProspEctive phArmacoeconomic cohoRt evaluation) Noninterventional Study Protocol

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Abstract

Background: Patients with multiple sclerosis (MS) require long-term therapy and have a wide variety of needs for health-related support. The efficacy and safety of MS therapy, as assessed by both clinicians and patients, are important parameters that need to be considered. However, few studies combine data on efficacy and safety outcomes with pharmacoeconomic data.

Objective: Here, we present the study design of the ProspEctive phArmacoeconomic cohoRt evaluation (PEARL), a prospective, multicenter, noninterventional cohort study on patients with relapsing-remitting MS (RRMS) treated with disease-modifying treatments (DMTs).

Methods: During a prospective observational phase of 24 months per patient, PEARL evaluated clinical and patient-perceived efficacy and safety measures, as well as pharmacoeconomic data on RRMS patients treated with DMTs—interferon beta and glatiramer acetate. Measurements of the patients' perceptions included the assessment of patient-reported quality of life, treatment satisfaction, and compliance. The study was planned to include 1800 outpatients from 180 German neurological practices who had continuously been treated with an approved DMT for at least 30 days. The primary statistical analyses of the PEARL study will be descriptive. Particular focus will be on specific subgroups, such as patients who switched DMTs during therapy and patients with disease worsening or disease activity. Subgroups will be compared using stratified analyses.

Results: Data collection for PEARL started in September 2010 and ended in July 2013. As of July 2015, the study is completed and is currently being analyzed and written up.

Conclusions: PEARL is evaluating both the health status and resource utilization of RRMS patients treated with DMTs in Germany. The combination of pharmacoeconomic data with clinical and patients' self-perceived efficacy and safety outcomes will add useful information to the currently incomplete picture of the overall RRMS burden in Germany.

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KEYWORDS

multiple sclerosis; outpatient care; efficacy; safety; pharmacoeconomics; disease-modifying therapies

Introduction

Multiple sclerosis (MS) is an inflammatory, demyelinating disease of the central nervous system with considerable physical, treatment-related, and economic consequences. With a lifetime risk of one in 400, MS is potentially the most common cause of disability in young adults [1]. Most patients present with the relapsing-remitting form of MS (RRMS) [2], which means that relapses with exacerbating symptoms alternate with remissions [3].

Management of RRMS requires a multimodal approach comprising both the treatment of acute relapses by corticosteroids and the suppression of disease activity by disease-modifying treatments (DMTs), including interferon beta (IFN-beta) preparations and glatiramer acetate (GA) [4]. Although early initiation and consistent administration of DMTs have been shown to decrease relapse rate and disease worsening [5-10], MS remains an incurable and debilitating disease. Since the life expectancy of MS patients is similar to that of the general population [11], MS patients require long-term therapy along with a continuous monitoring of drug efficacy, safety, and patients' satisfaction to avoid treatment-related complications and to improve treatment compliance [12,13].

Due to the wide distribution of lesions and diffuse disease processes, RRMS patients not only suffer from disability, but also from concomitant symptoms, such as visual loss, cognitive impairment, fatigue, and depression [14,15]. Quality of life declines with disease worsening [16,17]. Consequently, RRMS not only imposes severe physical hardship, but also a considerable psychosocial burden on patients, their families, and society [16]. Due to both the diversity of these physical, psychological, and social consequences and disease manifestation at a young age, RRMS is one of the most costly neurological diseases. The mean annual cost per MS patient—RRMS and progressive forms—in Europe is estimated at €27,000, which translated into total costs of €14.5 billion in 2010 [18].

This economic burden can be divided into direct and indirect costs. Direct costs represent the value of resources consumed to diagnose, treat, and accommodate MS patients with their condition, involving costs for pharmaceuticals, inpatient and outpatient care, and additional therapies such as physiotherapy. Indirect costs arise from unemployment, premature retirement, reduced productivity, and impaired quality of life [17]. In early MS stages, direct costs, predominantly for DMTs, account for the largest share of costs. Indirect costs increase during later MS stages, when disability and MS-associated symptoms become advanced [19]. There is surprisingly little comparative data on how these costs are differentially influenced by the use of DMTs in the outpatient setting. Moreover, many data sources do not include DMTs [20-23] or ignore direct nonmedical, indirect, or informal care costs [24,25]. Therefore, a detailed description of the economic burden associated with RRMS, in combination with efficacy and safety data, would be of great value for clinicians and health care providers.

Here, we present the study design of the ProspEctive phArmacoeconomic cohoRt evaluation (PEARL), a prospective, multicenter, noninterventional cohort study in RRMS patients treated with IFN-beta or GA. The 24-month observational phase of PEARL aims at collecting clinical efficacy and safety data, as well as pharmacoeconomic data on RRMS patients treated with approved DMTs in daily outpatient practice in Germany.

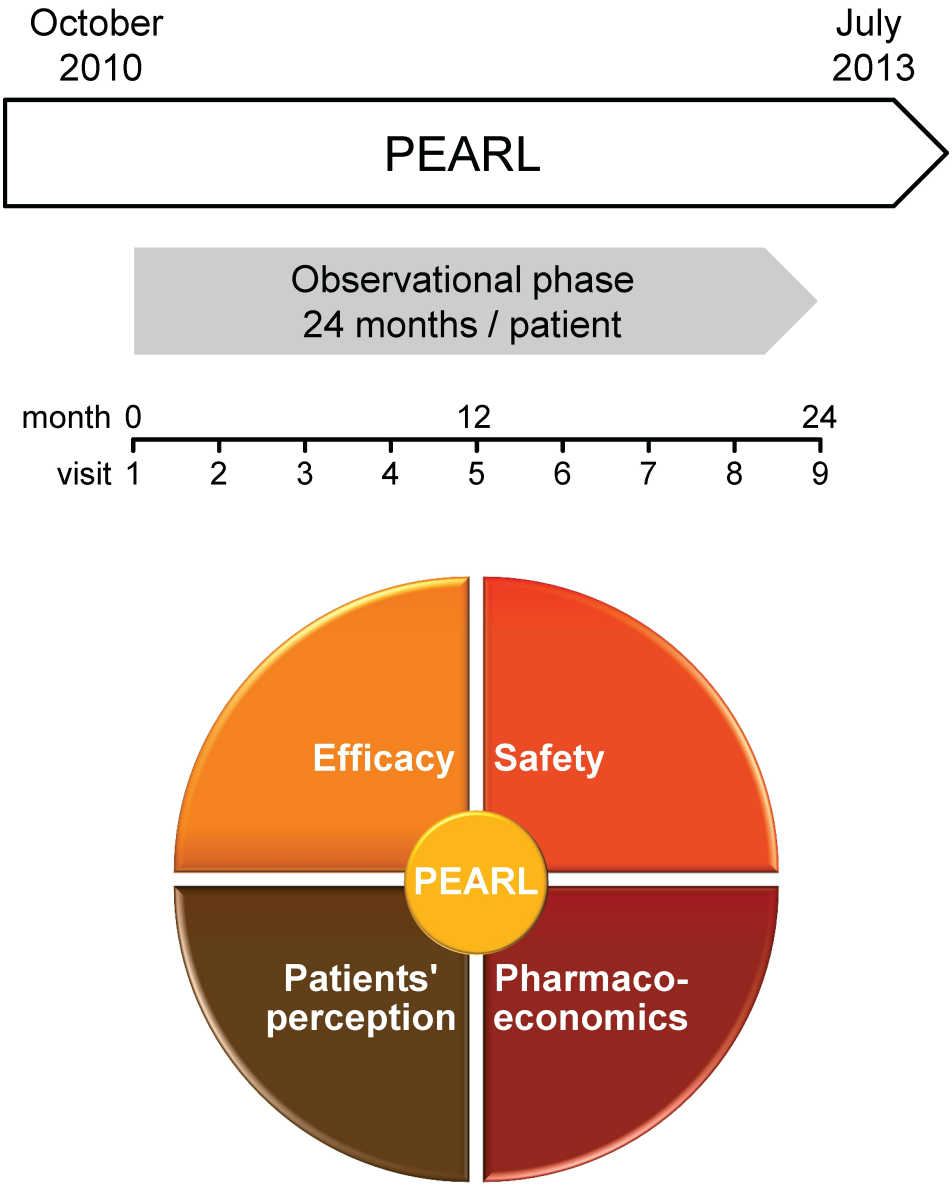
Methods

Study Design

PEARL is a prospective, multicenter, noninterventional cohort study of RRMS patients treated with IFN-beta or GA in daily outpatient care in Germany. The primary aim of this study was to collect and describe clinical and pharmacoeconomic data of RRMS patients treated with different DMTs. Clinical data comprised information on long-term efficacy and safety of DMTs, as assessed by clinicians and patients, and their effects on the patients' quality of life, treatment compliance, and treatment satisfaction. In this respect, data regarding the change between first-line DMTs and its impact on disease worsening were collected. Pharmacoeconomic data included information on both prescription of, and treatment with, DMTs and on resource utilization by RRMS patients. The study was designed to include 1800 patients from 180 neurological practices. The observational phase of 24 months per patient started in October 2010 and ended in July 2013 (Figure 1).

Approval from independent, local competent ethics committees was obtained, and the study was conducted in accordance with both the standards of the Association of Voluntary Self-Control of the Pharmaceutical Industry—Verein Freiwillige Selbstkontrolle für die Arzneimittelindustrie (FSA) [26]—and recommendations on the quality of noninterventional observational studies [27,28]. The study is registered as CNVF233ADE08 [29].

Figure 1. Timeline of PEARL, a noninterventional cohort study on relapsing-remitting multiple sclerosis (RRMS) patients treated with disease-modifying treatments (DMTs) in outpatient care. The study includes approximately 1800 RRMS patients from 180 neurological practices. Four major categories of data are evaluated during the observational phase of 24 months per patient (study visits 1–9). PEARL: ProspEctive phArmacoeconomic cohoRt evaluation.



Study Population

Patients with a diagnosis of RRMS were eligible if they had been continuously treated with an approved DMT—interferon (IFN) (Avonex, Betaferon, Extavia, or Rebif) or GA (Copaxone)—for at least 30 days, and if informed consent was given prior to study inclusion. There were no further selection criteria, except for the contraindications mentioned in the respective summaries of product characteristics [30-34]. The study protocol required therapeutic decisions to be made according to medical necessities, independently of study participation. To avoid selection bias, RRMS patients were enrolled in a consecutive order at each study center.

RRMS patients are normally treated in an outpatient setting. The rationale for the PEARL sample size was based on the estimated number of about 1300 neurologists in outpatient practice in Germany. Enrolling above 10% of the practices was deemed sufficient to obtain representative data. The selected number of 180 neurological practices/centers to be enrolled corresponds to 10-15% of total neurological practices and centers in Germany. Representativeness was further ensured by the regional distribution of participating practices and centers. The rationale to include 10 RRMS patients per practice was based on a practical anticipation of the practices' capabilities in study performance and their average potential to contribute patients based on expert advice. In 2010, approximately 200,000 statutory health-insured patients were diagnosed with MS—mostly RRMS and less progressive forms [35]. With an anticipated 1800 included RRMS patients, PEARL approximated 1% of German RRMS patients. With this and an observational phase of 24 months per patient, PEARL was deemed sufficient to obtain representative data on the most commonly prescribed DMTs in the outpatient setting.

Procedures

In accordance with routine outpatient care, participants were required to attend follow-up visits every 3 months, in addition to the first visit at baseline and the final visit after 24 months. Demographic and clinical data obtained from interviews, examinations, and medical records were transferred into the case report form (CRF) by the neurologist responsible. Patient questionnaires on patient-reported resource utilization, quality of life, treatment satisfaction, and compliance were completed by participants at regular visits in the presence of a health professional. Additionally, data on adverse events (AEs) were obtained directly from the patient. All patient-reported information was documented in the CRF by the neurologist responsible.

Data quality was ensured by validation checks at the time of data entry. Data were reviewed on an ongoing basis, and queries to the study sites were automatically initiated and followed. Data management was overseen by the clinical research organization responsible (Kantar Health GmbH). In study centers with three or more patients, on-site monitoring was performed by 2 months after the inclusion of the first patient. In 10% of study centers, observational data were checked against patient records after the end of observation.

Measures

Patient Characteristics

Table 1 shows the parameters to be assessed during study visits. After informed consent was given, patients formally entered the study. Demographic characteristics and patient histories, including prior and concomitant diseases and treatments, were assessed at baseline. Blood pressure and heart rate were evaluated at every visit beginning at baseline, and body weight was recorded at baseline and every 12 months.

RRMS-specific patient histories were retrospectively documented at baseline. These data included time since first symptoms, diagnosis and treatment of RRMS, the number of lesions in T2-weighted magnetic resonance imaging (MRI) as well as gadolinium-enhancing (Gd+) lesions, and the number and outcome of relapses within 12 months before study entry.

Concomitant RRMS treatments, medical and nonmedical, were documented at every visit beginning at baseline. Retrospective data on previous DMTs, including the time since start of treatment and the use of an auto-injector, were collected at baseline. Then, at every visit throughout the study, data on the current use of DMTs were documented, including the switch between DMTs and premature discontinuation of medication, as well as the reasons for discontinuation.

Clinical Outcome

The anamnestic number and outcome of MS relapses since the last study visit were documented at each study visit, and cerebral lesions were documented if MRI scans were available. To assess the overall clinical impression of functional impairment and disability, the Clinical Global Impression (CGI) scale [36] and the Kurtzke Expanded Disability Status Scale (EDSS) [37] were evaluated at baseline and every 3 and 6 months, respectively.

Adverse Events and Adverse Reactions

Occurrences of AEs, adverse drug reactions (ADRs), serious AEs (SAEs), and serious ADRs (SADRs) were evaluated by investigators at every study visit. AEs were defined as unfavorable and unintended signs or symptoms, complications, and changes of the patients' conditions during the observational phase, irrespective of relation to treatment. SAEs comprise life-threatening or fatal events, events requiring inpatient hospitalization or prolongation of hospitalization, events leading to major incapacity, persistent or significant disability, and congenital anomaly at birth, as well as events that are otherwise medically significant. The causality of any reported ADR and SADR was categorized as *certain*, *probable*, *possible*, *not assessable*, or *missing*.

Patients' Perceptions of Outcome

Patients' perceptions of DMT effectiveness was assessed by questionnaires on disability and quality of life. Patients' perceptions of disability were scored by means of the UK (Guy's) Neurological Disability Scale (UKNDS) [38] at baseline and every 12 months. Patients' perceptions of their quality of life were documented at baseline and every 6 months by using both the European Quality-of-Life Questionnaire (EQ-5D) [39] and the Patient-Reported Outcome Indices for MS quality-of-life (PRIMUS-QoL) and activity (PRIMUS-A) subscales [40]. Treatment satisfaction and treatment compliance were evaluated at baseline and every 3 months. Treatment satisfaction was assessed by the Treatment Satisfaction Questionnaire for Medication (TSQM-9) [41]. The compliance questionnaire focused primarily on whether, when, and how long DMT was eventually discontinued.

Table 1. Data obtained during PEARL[a] study visits.

Data obtained	Data obtained at various time points (X=data were obtained)		
	Baseline (Month 0)	Follow-up	Last visit (Month 24)
Patient characteristics			
Demographic data and patient history	X	N/A[b]	N/A
Heart rate, blood pressure	X	Every 3 months	X
Weight	X	Every 12 months	X
MS[c] history	X	N/A	N/A
Concomitant nonmedical MS treatments	X	Every 3 months	X
Prior and concomitant diseases and treatments	X	N/A	N/A
DMT[d] at baseline	X	N/A	N/A
Premature discontinuation of DMT	N/A	Every 3 months	X
Switch of DMT	X	Every 3 months	X
Efficacy			
MS relapses since previous visit	N/A	Every 3 months	X
MRI[e] lesions	N/A	If available	N/A
Kurtzke EDSS[f]	X	Every 6 months	X
CGI[g] scale	X	Every 3 months	X
Safety			
AE[h], ADR[i], SAE[j], and SADR[k]	X	Every 3 months	X
Patients' perceptions of outcome			
UKNDS[l]	X	Every 12 months	X
EQ-5D[m]	X	Every 6 months	X
PRIMUS-A[n] and PRIMUS-QoL[o]	X	Every 6 months	X
TSQM-9[p]	X	Every 3 months	X
Compliance patient questionnaire	X	Every 3 months	X
Pharmacoeconomic data			
Patient resource questionnaire	X	Every 3 months	X
Practice questionnaire	N/A	Once	N/A

[a]PEARL: ProspEctive phArmacoeconomic cohoRt evaluation.
[b]N/A: not applicable.
[c]MS: multiple sclerosis.
[d]DMT: disease-modifying treatment.
[e]MRI: magnetic resonance imaging.
[f]EDSS: Expanded Disability Status Scale.
[g]CGI: Clinical Global Impression.
[h]AE: adverse event.
[i]ADR: adverse drug reaction.
[j]SAE: serious adverse event.
[k]SADR: serious adverse drug reaction.
[l]UKNDS: UK (Guy's) Neurological Disability Scale.
[m]EQ-5D: European Quality-of-Life Questionnaire.

[n]PRIMUS-A: Patient-Reported Outcome Indices for Multiple Sclerosis activity subscale.

[o]PRIMUS-QoL: Patient-Reported Outcome Indices for Multiple Sclerosis quality-of-life subscale.

[p]TSQM-9: Treatment Satisfaction Questionnaire for Medication.

Pharmacoeconomic Data

Pharmacoeconomic data obtained in this study are primarily based on the analysis of the patient resource questionnaire, which was to be completed by patients at every visit beginning at baseline. In this questionnaire, several demographic data such as marital status, education, employment, and status of health and long-term care insurances were assessed by multiple-choice items. Additionally, participants were asked about their responsibility for relatives and the extent of concerns arising thereof on a 10-point Likert scale ranging from 0 (*not concerned at all*) to 10 (*maximally concerned*). The impact of RRMS on work productivity was assessed by *yes/no* questions combined with free-text fields asking about sick leaves and workplace changes. Patients were then asked to assess their productivity using another 10-point Likert scale ranging from 0 (*not affected by MS*) to 10 (*completely affected*).

Yes/no questions combined with free-text fields asked participants to state their financial expenditures on RRMS-specific and other medications, treatments, and devices. Patients were asked to specify outpatient therapies and medical consultations. Additional free-text fields asked for inpatient and outpatient care caused by MS relapses. Another question then asked if patients participated in nurse support programs such as EXTRACARE [42].

In addition to the patient-resource questionnaire, a practice questionnaire was completed once by each participating practice or center. This questionnaire collected information on the practice infrastructure and asked physicians and MS nurses for the number of patients treated and the time spent for diagnosis, therapy initiation, follow-up examinations, and advice. It further asked physicians to state the most important aspects mentioned in patients' interviews. The questionnaire then assessed the physicians' perceived treatment compliance and treatment satisfaction of RRMS patients and, additionally, their presumed underlying factors.

Statistical Analyses

All pharmacoeconomic, safety-, and effectiveness-related analyses will be performed for the full analysis set of patients, comprising patients who fulfilled all inclusion criteria mentioned above and attended at least one follow-up visit. In each analysis, the number of patients with missing data will be separately presented.

Primary statistical analyses are descriptive statistics such as mean, standard deviation, minimum, median, maximum, 25th and 75th percentiles, and number of nonmissing values. The descriptive statistical analysis will be used to summarize continuous variables, which will be additionally categorized in a clinically meaningful way. Frequency tables with absolute and relative frequencies will represent categorical data. For single efficacy parameters, such as CGI and EDSS scores as well as number of MS lesions, changes will be expressed as difference from baseline or summarized in shift tables. Subgroups defined by MS relapse will be compared using stratified analyses. For analyses, the statistical software SAS version 9.3 (SAS Institute Inc) will be used.

Results

Data collection for PEARL started in September 2010 and ended in July 2013. As of July 2015, the study is completed and is currently being analyzed and written up.

Discussion

Principal Findings

In this paper, we describe the study protocol of PEARL, a prospective, multicenter, noninterventional cohort study on RRMS patients treated with DMTs in daily outpatient practice in Germany. The aim of this study was to collect clinical and pharmacoeconomic data of a representative cohort of 1800 RRMS patients treated with different DMTs under routine outpatient conditions. The data therefore includes efficacy and safety-related outcome parameters as assessed by clinicians and patients, as well as pharmacoeconomic data of RRMS patients, including information on employment, work productivity, and resource utilization.

When assessing efficacy and safety, PEARL placed emphasis on the patients' perceptions. The patients' perceptions of their disability and health-related quality of life might differ from those of the attending physicians [43,44]. Therefore, evaluating the patients' perspectives is meaningful in improving the quality of treatment and health care services [12]. We further collected data on the frequency and reasons why patients switch DMTs and, moreover, assessed treatment satisfaction of RRMS patients. This information is clinically relevant because higher treatment satisfaction correlates with higher treatment compliance [13]; in turn, better compliance correlates with improved outcomes. It has been shown that the number of relapses can be reduced, and functional and cognitive abilities as well as quality of life can be improved, when RRMS patients comply with treatment in the long term [45,46]. Adherence to treatment has been demonstrated to reduce the risk of MS relapses and disability [47]. Since adherence rates in clinical trials seem to be higher than in routine care, the detailed assessment of treatment satisfaction and compliance in a real-world setting is necessary [48].

RRMS is obviously associated with significant economic burdens. Direct and indirect costs increase during relapses and disease worsening [49]. If new therapies are able to reduce relapses, delay accumulation of disability, relieve symptoms, and allow an acceptable quality of life, the overall economic burden on individuals and society might be reduced. Furthermore, the savings in indirect costs might outweigh direct costs of treatment. However, the current state of research in this area is subject to several uncertainties and data gaps. For example, most studies on the economic aspects of MS had been

conducted before DMTs were established as standard treatment, and results may no longer apply [20-23]. Moreover, results of other cost studies might not be representative because of the selection criteria applied. Our study therefore evaluated pharmacoeconomic information in combination with efficacy/safety data of different DMTs currently used in routine outpatient care. Except for contraindications to DMTs and the requirement that informed consent be obtained, no selection criteria were applied in our study to achieve the best possible representativeness.

Conclusions

The results of the PEARL study will add useful information to the currently incomplete data on the health status and resource utilization of RRMS patients treated with IFN-beta or GA in routine outpatient care in Germany. From both the economic and the clinical points of view, the results of PEARL will provide further insight into the health care costs and benefits of RRMS therapy and might support health care providers who are seeking ways to offer more cost-effective care.

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Conflicts of Interest

Stefan Viktor Vormfelde and Sonja Ortler are employees of Novartis Pharma GmbH, Nuremberg, Germany. Tjalf Ziemssen has served on scientific advisory boards, and has received scientific grants and speaker honoraria from Bayer, Biogen Idec, Genzyme, TEVA, Merck Serono, and Novartis.

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Abbreviations

ADR: adverse drug reaction
AE: adverse event
CGI: Clinical Global Impression
CRF: case report form
DMT: disease-modifying treatment
EDSS: Expanded Disability Status Scale
EQ-5D: European Quality-of-Life Questionnaire
FSA: Freiwillige Selbstkontrolle für die Arzneimittelindustrie
GA: glatiramer acetate
Gd+: gadolinium enhancing
IFN: interferon
IFN-beta: interferon beta
MRI: magnetic resonance imaging
MS: multiple sclerosis
N/A: not applicable
PEARL: ProspEctive phArmacoeconomic cohoRt evaluation
PRIMUS-A: Patient-Reported Outcome Indices for Multiple Sclerosis activity subscale
PRIMUS-QoL: Patient-Reported Outcome Indices for Multiple Sclerosis quality-of-life subscale
RRMS: relapsing-remitting MS
SADR: serious adverse drug reaction
SAE: serious adverse event
TSQM-9: Treatment Satisfaction Questionnaire for Medication
UKNDS: UK (Guy's) Neurological Disability Scale

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Original Paper

TOGETHER Project to Increase Understanding of the HIV Epidemic Among Sub-Saharan African Migrants: Protocol of Community-Based Participatory Mixed-Method Studies

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Abstract

Background: Sub-Saharan African Migrants (SAM) are the second largest group affected by HIV/AIDS in Belgium and the rest of Western Europe. Increasing evidence shows that, more than previously thought, SAM are acquiring HIV in their host countries. This calls for a renewed focus on primary prevention. Yet, knowledge on the magnitude of the HIV epidemic among SAM (HIV prevalence estimates and proportions of undiagnosed HIV infections) and underlying drivers are scarce and limit the development of such interventions.

Objective: By applying a community-based participatory and mixed-methods approach, the TOGETHER project aims to deepen our understanding of HIV transmission dynamics, as well as inform future primary prevention interventions for this target group.

Methods: The TOGETHER project consists of a cross-sectional study to assess HIV prevalence and risk factors among SAM visiting community settings in Antwerp city, Belgium, and links an anonymous electronic self-reported questionnaire to oral fluid samples. Three formative studies informed this method: (1) a social mapping of community settings using an adaptation of the PLACE method; (2) a multiple case study aiming to identify factors that increase risk and vulnerability for HIV infection by triangulating data from life history interviews, lifelines, and patient files; and (3) an acceptability and feasibility study of oral fluid sampling in community settings using participant observations.

Results: Results have been obtained from 4 interlinked studies and will be described in future research.

Conclusions: Combining empirically tested and innovative epidemiological and social science methods, this project provides the first HIV prevalence estimates for a representative sample of SAM residing in a West European city. By triangulating qualitative and quantitative insights, the project will generate an in-depth understanding of the factors that increase risk and vulnerability for HIV infection among SAM. Based on this knowledge, the project will identify priority subgroups within SAM communities and places for HIV prevention. Adopting a community-based participatory approach throughout the full research process should increase community ownership, investment, and mobilization for HIV prevention.

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KEYWORDS

HIV; HIV prevention; sub-Saharan African migrants; HIV prevalence; HIV risk factors; mixed methods; community-based participatory research; oral fluid; electronic questionnaire; surveys and questionnaires

Introduction

Background

"Know your epidemic, know your response," has become the directive of the Joint United Nations Programme on HIV and AIDS (UNAIDS) for intensifying HIV prevention [1]. Sub-Saharan African Migrants (SAM) are the second largest group affected by HIV/AIDS in Belgium [2,3]; however, knowledge on the population's HIV prevalence and underlying factors that shape the HIV epidemic in SAM communities is scarce. This limits the development of targeted primary prevention interventions. Such interventions have gained importance, since recent evidence shows that increasing proportions of SAM acquire HIV in their European host countries [4-8]. To address this knowledge gap and improve primary prevention for SAM, we developed the TOGETHER Project, which started in January 2012. The project's study protocol is presented in this paper.

HIV in Belgium's African Communities: "What Do We Know?"

In Belgium, 27% (n=230) of the newly reported HIV diagnoses in 2013 were in individuals of sub-Saharan African origin [3]. Since in 24% of all reported cases, data on the country of origin was missing, it is assumed that the overall number of new HIV diagnoses among SAM might be underestimated. Yet, it is clear that as communities of SAM are small (1.6% of the Belgian population), HIV disproportionately affects them.

Reported characteristics of newly diagnosed SAM in Belgium are in line with the generalized epidemic in sub-Saharan Africa. In 2013, the majority (64%) were women, heterosexual contact was the main transmission mode (89%), and most (78%) were diagnosed between 20 and 45 years [3]. Patients originated from 31 different countries, the largest groups coming from Cameroon (18%), Democratic Republic of the Congo (12%), and Guinea (9%) (personal communication Sasse, 2015). As in other European countries [2], HIV infections among SAM were usually diagnosed late. Of newly diagnosed SAM, 50% were late presenters, that is, their CD4 cell count was < 350/ml at the time of first diagnosis in Belgium [3]. According to CD4 cell decline simulations, it takes an estimated period of about four years from seroconversion for the CD4 count to reach 350 [9]. During this time, diagnostic opportunities may be missed. However, HIV diagnosis does not automatically translate into direct linkage to care among SAM [10,11]. Denial, coming to terms with diagnosis, not knowing where to go, feeling well and having no symptoms, HIV stigma and discrimination, and fear of medication prevent SAM from accessing care immediately after diagnosis [11].

Late diagnosis and delayed initiation of care not only affect disease prognosis [12], life expectancy [13], and health care costs [14-16], but also lead to an increased risk for onward HIV transmission, due to the prolonged period of unawareness of HIV status and continued high viral load of those not treated [17,18].

Belgian surveillance data show that 10.4% of all SAM diagnosed in 2013 report having acquired HIV in Belgium. Yet, this is based on the physician's assessment at diagnosis and data are missing for 30% of cases [3]. Evidence from other EU countries suggests that this might be an underestimation. An Italian study among newly arrived immigrants first suggested that HIV infection had been more often acquired in the host country than previously estimated [6]. A study among newly diagnosed Africans in London specified that a quarter to a third of all HIV-positive Africans residing in the UK, and nearly half of HIV-positive African men who have sex with men (MSM), were likely to have acquired HIV in the UK [5]. Mathematical modeling applied to the UK's national HIV-diagnosis data of heterosexuals born abroad suggests an increasing trend. While in 2004 an estimated 24% had acquired HIV after migration in the UK, this rose to 46% in 2010 [4], which accounts for an absolute increase of 16.5%. Preliminary results of applying the same mathematical model to Belgium's national HIV surveillance data suggested that among patients newly diagnosed in 2011, 28% (IQR 24%-33%) of non-Belgium born heterosexuals and 39% (IQR 32%-47%) of non-Belgium born MSM could have acquired HIV in Belgium [7]. The close-knit sexual networks of SAM [19], combined with structural vulnerability related to the migration context [20,21], may contribute to an increased risk for HIV infection among SAM residing in Belgium.

Knowledge Gaps for Effective HIV Prevention

HIV prevention comprises the continuum of primary prevention, promotion of HIV testing and counselling, and "positive health, dignity, and prevention," which includes the prevention of HIV transmission. Yet, in Belgium [22] and its neighboring countries [23], prevention at the community level in the last decade focused mainly on the promotion of HIV testing and early linkage to care. Deeper understanding of SAM's increased risk of being diagnosed late and the barriers to and facilitators of HIV-testing uptake [2,24-26] led to the development of multiple HIV-testing promotion strategies [14-16].

The prevention of new HIV infections, or primary prevention, among SAM has not been made priority because traditionally the HIV epidemic in the African diaspora in Europe was understood to be imported [27]. Evidence of SAM acquiring HIV after migration highlighted the need for tailored primary prevention interventions [4,5]. However, a number of gaps in in-depth understanding of transmission dynamics still exist. First, HIV prevalence estimates for a representative sample of the SAM communities in Europe are lacking, complicating HIV risk assessment and awareness raising in the communities. SAM have mostly formed subgroups in studies on HIV prevalence

among other target groups, such as immigrant female sex workers [28], MSM [29], recently arrived migrants [6], and immigrants [30-34]. Only the MAYISHA II study, conducted in 2004 among 1359 black Africans in London, Luton, and the West-Midlands, provided HIV prevalence estimates of 14% [35]. Using a convenience sample of recruitment sites, this study did not include a representative sample, and subsequently, oversampling of HIV-positive individuals could not be excluded [4,5].

Second, estimates of the proportions of SAM with undiagnosed HIV are lacking. In Europe, one third of persons living with HIV are assumed to be unaware of their HIV status [21]. Some of the above-mentioned HIV prevalence studies provided indications that this might be higher among SAM; for example, MAYISHA II found that 66% of study participants with a positive test result did not report their HIV status on the questionnaire [35].

Third, previous research mainly focused on SAM's individual knowledge, attitudes, and practices related to sexuality and HIV-preventive behavior, thus underestimating the social, cultural, religious, and migration-related contexts that increase vulnerability with respect to HIV [36]. Studies from the UK and the Netherlands underlined SAM's preference for the heterosexual, monogamous standard [35]; yet, high number of partners (lifetime [37] and past year [30,35]), concurrent relationships, and having sex when traveling to home countries [30,34,37] were frequently reported, especially among men. This increased individual sexual risk behavior was shown to be linked to higher rates of sexually transmitted infections among SAM [34,37-39]. Several studies showed that SAM's condom use was relatively high in comparison with the West-European population, but low considering their potential risk [40]. Africans were reported to believe that risk could be avoided by carefully choosing their sexual partners [40], to believe that condoms were associated with infidelity and reduced sexual pleasure, and to perceive condoms as inappropriate for long-term relationships [39,41-43]. Although these studies were useful to identify individual sexual risk patterns, they paid little attention to diversity among subgroups [36] and contextual factors influencing sexual behavior, and thus may be of limited use to develop and implement effective campaigns aiming to reduce HIV infection risk [44,45].

SAM Communities in Antwerp City

Although small in numbers, SAM communities are characterized by a high degree of heterogeneity due to diverse ethnic and cultural backgrounds, migration patterns and residence statuses, educational and socioeconomic backgrounds, and religious beliefs [22,45]. Of the 175,000 SAM officially living in Belgium [46], about 17,400 (10%) reside in Antwerp city (according to data obtained from the City of Antwerp; email communication from 2012 May 23). These numbers include SAM who obtained Belgian nationality, second-generation Belgian-born children of SAM parents, registered migrants, and SAM whose residence procedure is pending. SAM of undocumented legal status are absent from these statistics. Almost half (47%) of SAM in Antwerp city originate from 3 countries: the Democratic Republic of the Congo (18.8%), Ghana (17.5%), and Nigeria (10.7%). Apart from these 3 main nationalities, 43 other nationalities are living in this area. In spite of their heterogeneity, SAM communities are fairly homogeneously organized. For many SAM, their nationality and/or ethnicity shape their social life. As new migrants, they depend on the social support of their compatriots to become settled [47]. As established migrants, they engage in social and cultural networks and use them to look for marriage and sex partners [30]. Although these ethnic networks can be described as tight, they are not ethnically segregated. Different ethnic groups mingle at commercial and social venues and events. To reach different communities with HIV prevention activities, partnerships with leaders (ie, of sociocultural or spiritual organizations and owners of commercial settings) are essential [48-50].

Many SAM live in socioeconomically vulnerable and legally unstable conditions. Together with prevalent HIV-related stigma and culturally grounded taboos on sexuality, this translates into little demand for HIV prevention [50,51] and increased risk for HIV acquisition [48].

Methods

Study Objectives

The TOGETHER study's overall aim was to increase the communities', researchers', and policymakers' in-depth understanding of the dynamics of the HIV epidemic among SAM, to improve primary prevention interventions. This translated into the following objectives:

1. To assess the HIV prevalence and proportion of undiagnosed HIV infections among SAM socializing in community settings in Antwerp city.
2. To identify individual, community-level, and structural risk factors for HIV infection among SAM.
3. To identify priority settings and groups for future primary HIV prevention interventions.
4. To increase community ownership, involvement, and mobilization for HIV prevention.
5. To develop policy recommendations to improve HIV prevention for the target group of SAM.
6. To assess the feasibility and acceptability of community-based participatory research on HIV prevalence in SAM communities and adopted research tools.

Overall Study Design

To meet these objectives, the TOGETHER Project applied mixed methods and a community-based participatory research approach (CBPR) [52]. The main study was a cross-sectional community-based bio-behavioral survey on HIV prevalence and HIV risk factors among SAM visiting community settings in Antwerp city (referred to as the "HIV prevalence study" in this article). To inform its design, 3 formative substudies were conducted. First, a social map of SAM community settings in Antwerp city was developed, applying an adaptation of the PLACE Method [53,54]. Second, factors that increase SAM's risk of HIV infection were assessed using a multiple case study design. The third substudy assessed the acceptability and feasibility of using oral fluid collection devices for HIV testing

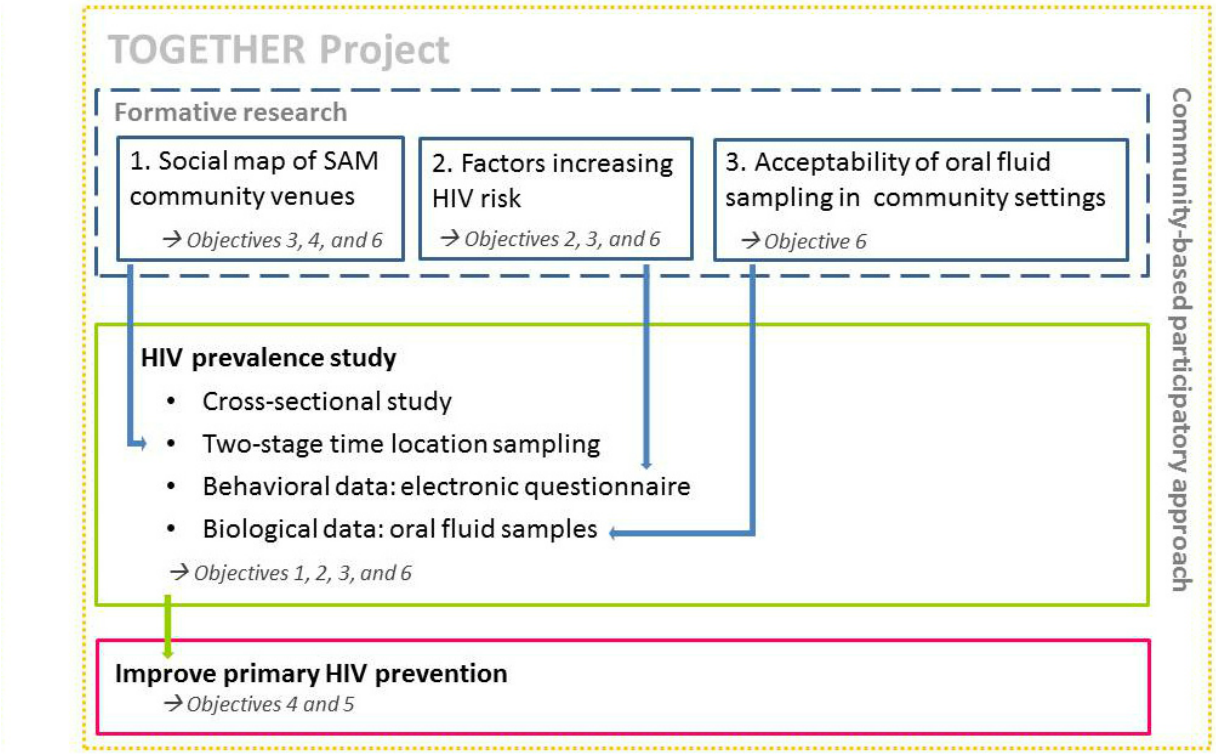
in community venues through participatory observations, including informal interviews (see Figure 1).

For the different study components, separate study protocols were developed and ethical approval was obtained from the Institutional Review Board of the Institute of Tropical Medicine and the ethical committee of the University Hospital Antwerp.

Community-Based Participatory Approach

To account for the heterogeneity of Antwerp's SAM and to ensure that the study methods and tools were acceptable for all subgroups (objective 6), we chose a community-based participatory approach [52]. This methodology also allows for increasing community ownership, involvement, and mobilization for HIV prevention (objective 4) throughout the research process. In practice, community members are involved in all steps of the project, from conceptualization, to data collection, to development of new interventions and policy recommendations. We established collaborative partnerships both with research experts and communities [55], by engaging a team of community researchers (CRs) and setting up a community advisory board (CAB). The CAB hosted both leaders of African organizations and a multidisciplinary group of professionals. The CRs were 9 lay community members, who were trained at the start of the project and continuously coached throughout. To reflect the communities' diversity, the CR team was diverse in its composition with respect to gender, age, origin, residence status, employment status, and HIV status. In line with the Greater Involvement of People living with HIV/AIDS (GIPA) principles [56], we actively recruited SAM living with HIV for the CRs team.

Figure 1. Outline of the TOGETHER project.



Formative Research

Formative Study 1: Social Mapping of Sub-Saharan African Community Venues in Antwerp City

This study ran from June 2012 until June 2013 and had the triple objective of (1) determining the sampling frame for the HIV prevalence study, (2) identifying priority settings for future HIV prevention, and (3) increasing communities' ownership of HIV prevention.

To account for the heterogeneity of the SAM communities and ensure inclusion of hidden subpopulations, for example, SAM of undocumented status or MSM, we chose to employ the systematic approach of the PLACE Method (Prioritising Local AIDS Control Efforts) [53,54]. Guided by epidemiological theories, the PLACE Method has been widely used to monitor and improve AIDS prevention program coverage in areas where HIV transmission is most likely to occur, for example, by identifying venues where people meet sexual partners. It is a 5-step method adopted for surveillance studies, intervention design, programmatic up-scaling, and community mobilization [53,54,57-59]. We adapted the method to better meet our objectives by including steps 1 to 3, while replacing steps 4 and 5 with the HIV prevalence study (see below).

In step 1, "identifying a priority prevention area," Antwerp city was selected based on demographic and epidemiological aspects: 22% of all SAM in Flanders live in Antwerp city [60] and, according to data available at the time, in 2010 43% of all newly diagnosed SAM in Flanders lived in Antwerp province. In step 2, "community informants" were interviewed, that is, adults who are knowledgeable about the community. Participants were asked where SAM socialize, and where they meet new sexual

partners. We collected the locations of various publicly accessible meeting places such as bars, churches, shops, hair salons, asylum centers, a public library, parks, streets, squares, events, and festivities of African organizations. A convenience sample of community informants with different behavioral and sociodemographic characteristics, and of different professions, and community leaders of different origins, residing in different city districts, were interviewed to assure representativeness. After reaching saturation, duplications were removed from the compiled inventory of places and a consolidated list was generated. In step 3, all venues, areas, and events on the list were visited for a verification interview. Structured electronic questionnaires (SurveyToGo) were used to assess settings' activities, characteristics of their visitors (origin, gender, etc.), busiest days and times, and existing HIV prevention programs, as well as to collect information on additional settings.

Formative Study 2: Assessing Factors that Increase Sub-Saharan African Migrants' Risk of HIV Infection: A Multiple Case Study

The second formative study contributed to the second overall project objective (ie, assessing individual, community-level, and structural risk factors). This study's first phase took place between April 2013 and December 2013. The 3 specific objectives were: (1) informing the development of a structured questionnaire for the HIV prevalence study, (2) qualitatively contextualizing the findings of the HIV prevalence study, and (3) informing the development of future HIV prevention interventions. Study participants were SAM living with HIV, who were unique cases to retrospectively identify multi-level risk and vulnerability factors.

We conducted an empirical inquiry investigating a phenomenon within its real-life context, using multiple sources of evidence [61]. In our study, individual HIV positive SAM were the single object of a case. For each case, we triangulated findings from 3 data collection methods: life history interviews, timelines, and patient files. Per case, at least two life history interviews were conducted. Since chronology, sequencing of events, and context [62] are important to understand the setting in which HIV infection occurred, *timelines* were developed during these interviews. Timelines are visual depictions of an individual's life events in chronological order that may include interpretations of these events [62]. We followed the timeline approach as described by Adrianson (2012), in which the drawing of the timeline is a collaborative effort shared by the interviewer and the interviewee, with the drawing forming the basis of the interview [63]. All interviews were conducted by a single interviewer (JL) who adopted an unstructured interview approach. This informal, open-ended, flexible, and free-flowing way of interviewing enabled participants to define the properties of the interview and direct the interview into areas that they saw as relevant [64]. The interview themes were the following: life story and events, migration (eg, pull and push factors, trajectory, immigration procedures), sexual and partner relationships, health-seeking behavior and coping styles and emotions (eg, effect of events on psychological well-being, coping with HIV, stigma and discrimination), social embeddedness (eg, social network, family structure and support, social exclusion) and livelihood (eg, financial situation, housing). When the natural flow of the first interview did not spontaneously generate sufficient data on these themes, a focused approach was taken in the second and eventual follow-up interviews. Data from the life history interviews were triangulated with data from the patient files. This reduced potential limitations of life history interviews (eg, subjective distortion), enriched the findings, and increased internal validity within the cases.

In the first study phase, a convenience sample of SAM living with HIV was recruited through physicians and nurses of the HIV clinic and facilitators of an HIV support group for SAM. To arrive at a representative sample of the patient population of sub-Saharan African origin, in the second phase, with a start date of April 2016, we switched to purposive sampling.

All participants in the first study phase were consenting adults who received their HIV diagnoses between 6 months and 10 years ago, who were assessed by health care providers as being psychologically stable and were followed up by a social nurse at the time of the interview. The latter was important to assure linkage to psychosocial care in case the interviews evoked emotional upheaval. The same rationale led to significant attention being paid to the informed consent procedure. Prior to the first interview, the study's rationale, objectives, procedure, and confidentiality measures, and participants' rights, benefits, and disadvantages were discussed extensively with the participant, before the informed consent form was signed. During this process, we asked for explicit approval to consult the participants' patient files. For follow-up interviews, the procedure was repeated and verbal informed consent was obtained. As a token of appreciation, participants received an incentive of €25 for each interview.

To ensure participants' anonymity and confidentiality, all data were coded and stored in a password-protected folder. All data were uploaded to NVivo 10 and a first *within-case analysis* was conducted. Data analysis adopted an inductive approach [65]. Triangulating the different data sources, the specific study questions were answered for each single case. Next, a cross-case analysis will be conducted at the end of the second study phase, to identify general factors that increase SAM's vulnerability to HIV infection and facilitators of and barriers to behavior change.

Formative Study 3: Acceptability and Feasibility of Outreach HIV Testing Using Oral Fluid Collection Devices

For the HIV prevalence study, we opted for collecting oral fluid samples to determine HIV status, since reluctance toward blood taking is known to limit HIV testing uptake among SAM [66]. Although oral fluid collection devices had been used in comparable studies [30,35], none took place in Belgium; thus, their acceptability in SAM community settings was unknown. Therefore, an acceptability study was conducted between December 2012 and June 2013 within the framework of another HIV testing intervention named "swab2know."

This intervention of the Institute of Tropical Medicine offered free oral fluid HIV tests (Oracol device, Malvern Medical Developments, Worcester, UK) in community settings of two target groups (MSM [67] and SAM) in Antwerp city. The HIV

testing sessions for SAM were organized in collaboration with community leaders and included group counseling and testimony of an HIV-positive community member. If participants decided to test, they could choose to collect their result a week later via a secured website or face-to-face consultation at a low-threshold HIV testing center (ie, focusing on high-risk groups such as SAM). To assess the feasibility and acceptability of this intervention, including the specific sampling method for SAM, two social scientists (JL and FN) conducted participant observations [68] at 10 HIV testing sessions. Besides observation, informal interviews were conducted with testers, nontesters, and the intervention team. Field notes were coded using NVivo 10 using a data-driven code book and analyzed following inductive analysis principles [65].

Community-Based Survey on HIV Prevalence and HIV Risk Factors Among Sam Visiting Community Venues in Antwerp City

The primary objective of this cross-sectional study, which ran from December 2013 to August 2014, was to determine HIV prevalence among SAM socializing in community settings in Antwerp city (objective 1 of the TOGETHER Project). Secondary objectives were: (1) Identifying the individual, community-level, and structural risk factors for HIV infection among SAM; and (2) Identifying priority settings for future HIV prevention interventions (project objectives 2 and 3, respectively).

Sample Size

HIV prevalence was the primary outcome measure. The sample size was calculated using an anticipated HIV prevalence of 4%, a required precision of 2% for the 95% confidence intervals, and a cluster sampling design effect of 2. This resulted in a required sample size of 714 SAM.

Sampling Method

A 2-stage time location sampling (TLS) was adopted. TLS takes advantage of the fact that some hard-to-reach populations tend to gather or congregate at certain types of locations [69]. The list of settings established in formative study 1 was the sampling frame, from which a 2-stage cluster probability sample was selected. At the first level of sampling, 51 clusters, or sites, were randomly selected from the list with a probability proportional-to-size. When a selected site was not available (eg, refusal of the bar owner, closure of the site, site moved out of study area), this was noted and the next site on the list was approached. The second level of sampling included the random selection of 14 study participants from each cluster.

To be eligible, potential study participants had to self-identify as belonging to the SAM communities, be 18 years or older, agree to answer the behavioral questionnaire, donate an oral fluid sample, and be willing and able to provide written informed consent. Prior participation in the prevalence study was an exclusion criterion.

Measures

The study combined biological and behavioral measures. To measure the study's primary outcome, HIV prevalence, oral fluid samples were collected and tested for HIV antibodies in the AIDS Reference Laboratory. These samples were linked through a unique code to an anonymous behavioral questionnaire. This instrument included questions on participants' socio-demographic and economic background, migration and mobility background, health-seeking behavior, HIV-testing behavior, sexual and relational history (last year and lifetime), attitudes towards condom use, actual condom use, and level of assistance needed to complete the questionnaire. The structured electronic questionnaire was developed based on the findings of formative study 2, consultation of available questionnaires from comparable studies [39,42], and input from the CRs and CABs. It was refined after cognitive piloting with 12 participants and the pilot sessions (see below).

Data Collection Procedures

Detailed study procedures were described in the Standard Operating Procedures (SOP) developed in collaboration with the CRs, CAB, and the AIDS reference laboratory. The SOP were refined after 2 pilots. At pre-arranged moments, a study team visited the selected sites and randomly selected 14 of the SAM present. When approaching potential participants, the CRs identified themselves and introduced the study's objectives and methodology, stressing the anonymous and voluntary nature of participation. To avoid self-exclusion of HIV-positive individuals, they explicitly mentioned that everybody was invited to participate, regardless of HIV status. Interested individuals were invited to a quiet area in the setting, if available. After discussing and signing the informed consent form, participants were asked to complete a structured anonymous electronic questionnaire on a tablet through SurveyToGo. To build confidence and ensure data rigor, participants first received a short tutorial on how to use the tablet. Here, special attention was given to the demonstration of how anonymity was guaranteed. Questionnaires were available in French, English, and Dutch, which are reference languages for most SAM. The preferred interview method was self-completion; however, assistance was offered if needed. By ethnically matching the CRs to the settings, translation of questions to a local language was possible, if preferred. The CRs were trained to offer assistance with sensitivity and respect to confidentiality.

After completing the questionnaire, the CR demonstrated the procedure of oral fluid collection using the collection device. Next, participants were asked to self-collect the sample. The sample was then linked with the informed consent form, questionnaire, and a letter explaining how to collect the results through a *unique code* (no personal identifying information was requested). If they wished, participants could collect their HIV test result by calling the study nurse and providing their unique code, age, and country of origin. When results turned out to be HIV positive, participants were invited to the HIV testing center for confirmation testing, counselling, and linkage to care.

Finally, participants were asked to provide information on their frequency of *attending* the study settings and comparable settings. This allowed the calculation of a weighting factor (see "data analysis"). As a token of appreciation, participants received free condoms, an information brochure on HIV testing, and €5. Those who refused participation received condoms and

the information brochure, but no financial compensation. Data on their characteristics and reasons for refusal were also collected, as well as the overall number of individuals present during the onsite data collection.

Laboratory Procedures

Within 7 days of collecting the sample, the AIDS reference laboratory of the Institute of Tropical Medicine performed the analysis according to a validated algorithm using oral fluid specimens [70]. First, samples were tested with a Genscreen HIV ½ v2 (BioRad). If reactive, a second HIV ELISA test, Vironostika HIV Ag/Ab (BioMérieux) was performed. Only participants with 2 reactive test results were considered to be HIV-infected. For all negative samples, the quality of the oral fluid samples was measured using an IgG ELISA quantification kit (Human Total IgG ELISA, Immunology Consultants Laboratory, Inc, Cat No: E-80G). Samples were considered valid for analysis and results of the HIV test were only reported when sufficient IgG was present in the sample. All other samples were considered nonvalid and excluded from analysis.

Data Analysis

Data from the questionnaires, attendance forms, laboratory data (HIV status), and HIV test result collection form were linked via the unique code, merged, and stored in an SPSS Statistics 22 (IBM) database. After data cleaning, statistical analysis was carried out. An analysis plan to take into account cluster sampling and a weighting factor [71] was developed using IBM SPSS Complex Samples 22. SAM who visited sites more frequently had a higher probability of selection in the study. Adjustment for this unequal selection probability was completed by calculating individual weights, based on the attendance information provided by the participant as provided on the venue attendance form. The first step of the data analysis was a univariate descriptive analysis of all outcome variables, stratified by gender, including HIV prevalence. Categorical variables were summarized by proportions and 95% confidence intervals. Nonnormally distributed quantitative data were described by median and interquartile ranges. During a second step, bivariate analysis was conducted by exploring potential determinants of HIV infection and HIV-risk taking behavior. Odds ratios were calculated to measure the association and statistical significance testing was done using a chi-square or *t* test. A multivariate analysis still has to be performed, using a logistic regression model constructed with all variables independently associated with HIV infection, sexual risk behavior, condom use, and testing behavior.

Results

Results have been obtained from 4 interlinked studies and will be described in future research.

Discussion

The TOGETHER Project was conceptualized to respond to the growing need for investing in targeted primary prevention interventions to reduce new HIV infections among SAM. Increasing evidence indicates that more SAM than previously assumed acquire HIV after migration to West European host countries [4-8]; however, in-depth understanding of individual, community-level, and structural factors that increase risk and vulnerability for HIV are lacking. This hampers the development and implementation of tailored responses. Applying a mix of empirically tested and innovative epidemiological and social science methods through a CBPR approach could potentially reduce this knowledge gap, while increasing community ownership, investment, and mobilization for prevention. Against this background, the project's objective was to generate the first HIV prevalence estimates for a representative sample of SAM residing in a Western European country and to yield an in-depth understanding of the multiple factors leading to HIV infection among SAM.

Our HIV prevalence study adopted methods successfully implemented in previous bio-behavioral surveys and cross-sectional studies [30,34,35,55], that is, combining anonymous biological data from oral fluid sampling with behavioral questionnaire data collected in community venues and public areas by ethnically matched volunteer interviewers. To obtain reliable HIV prevalence estimates representative of the diverse and heterogeneous SAM communities socializing in Antwerp city, we extended these methods with a 2-stage time location sampling approach. A limitation of the time location sampling is the difficulty in estimating the probability of missing an individual who never attends any of the venues listed. We adopted extensive formative research to develop an exhaustive social map of community settings covering all aspects of social life in order to limit that probability.

Study methods and tools were continuously refined based on the input of a CAB, CRs team, and the findings of two additional formative studies. To the best of our knowledge, this was the first time that such a carefully designed CBPR design had been adopted in Europe to assess HIV prevalence among a representative sample of SAM.

Due to practical limitations, the study setting was limited to Antwerp city. While Antwerp is the major city where SAM reside in Flanders, this implies that, although the HIV prevalence study will generate a sound estimation of HIV prevalence among SAM, the results cannot be generalized to Belgium or other West European countries. Different compositions of SAM communities in other cities have to be considered. For example, in Brussels 47% of SAM are of Congolese origin [46], while in the UK a vast majority come from Eastern and Southern Africa [72].

Adopting the CBPR approach as outlined is known to be challenging in terms of assuring skills of lay researchers, and safeguarding continuous motivation and data quality [52,73]. Our experience shows that the advantages outweighed these potential limitations. Adopting a CBPR approach improved access to hidden and hard-to-reach subpopulations, ensured acceptability of the study and its methods, built community ownership for HIV prevention, respected GIPA principles, and made the HIV epidemic real to individuals in the community, as was also shown by a comparable study among MSM in Antwerp city [74]. To generate this effect, we invested in sufficient training, individual coaching, and regular meetings

with the CRs and set up rigorous data quality assurance measures throughout the entire research process.

Ability to ensure community mobilization for HIV prevention may be challenged by time constraints. While some subgroups are stable communities, others are known to evolve rapidly, community venues are often unstable, and leadership fluctuates [22]. Therefore, the projects' results concerning priority places for prevention (objective 3) need constant follow-up (eg, to track where venues moved to, to assure contacts with new leaders).

With respect to the multiple-case study, it should be mentioned that our approach to identifying factors that increase SAM's vulnerability to HIV infection was innovative (ie, triangulation of data from life history interviews, lifelines with patient files). In particular, the use of lifelines was, according to our literature search, new to HIV research. These qualitative insights were valuable in the analysis and contextualization of the HIV prevalence study's findings. Together, they will form a solid basis for policy recommendations and the development of future HIV prevention interventions, once all study results are available.

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Conflicts of Interest

None declared.

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Abbreviations

CAB: community advisory board
CBPR: community-based participatory research
CRs: community researchers
GIPA: greater involvement of people living with HIV/AIDS

MSM: men having sex with men
PLACE: Prioritising Local AIDS Control Efforts
SAM: sub-Saharan African migrants
TLS: time location sampling

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Original Paper

Treatment of Internet Addiction with Anxiety Disorders: Treatment Protocol and Preliminary Before-After Results Involving Pharmacotherapy and Modified Cognitive Behavioral Therapy

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Abstract

Background: The growth of the Internet has led to significant change and has become an integral part of modern life. It has made life easier and provided innumerous benefits; however, excessive use has brought about the potential for addiction, leading to severe impairments in social, academic, financial, psychological, and work domains. Individuals addicted to the Internet usually have comorbid psychiatric disorders. Panic disorder (PD) and generalized anxiety disorder (GAD) are prevalent mental disorders, involving a great deal of damage in the patient's life.

Objective: This open trial study describes a treatment protocol among 39 patients with anxiety disorders and Internet addiction (IA) involving pharmacotherapy and modified cognitive behavioral therapy (CBT).

Methods: Of the 39 patients, 25 were diagnosed with PD and 14 with GAD, in addition to Internet addiction. At screening, patients responded to the MINI 5.0, Hamilton Anxiety Rating Scale, Hamilton Depression Rating Scale, Clinical Global Impressions Scale, and the Young Internet Addiction Scale. At that time, IA was observed taking into consideration the IAT scale (cutoff score above 50), while anxiety disorders were diagnosed by a psychiatrist. Patients were forwarded for pharmacotherapy and a modified CBT protocol. Psychotherapy was conducted individually, once a week, over a period of 10 weeks, and results suggest that the treatment was effective for anxiety and Internet addiction.

Results: Before treatment, anxiety levels suggested severe anxiety, with an average score of 34.26 (SD 6.13); however, after treatment the mean score was 15.03 (SD 3.88) (P<.001). A significant improvement in mean Internet addiction scores was observed, from 67.67 (SD 7.69) before treatment, showing problematic internet use, to 37.56 (SD 9.32) after treatment (P<.001), indicating medium Internet use. With respect to the relationship between IA and anxiety, the correlation between scores was .724.

Conclusions: This study is the first research into IA treatment of a Brazilian population. The improvement was remarkable due to the complete engagement of patients in therapy, which contributed to the success of the treatment from a behavioral perspective, and gave patients the confidence to continue to manage Internet use in their lives.

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KEYWORDS

Internet Addiction; Panic Disorder; Generalized Anxiety Disorder; Treatment; Cognitive Behavioral Therapy; Anxiety Disorders; Cognitive Therapy; Therapeutics

Introduction

Background

The rapid expansion of the Internet and its integration into modern life has led to far-reaching changes in our day-to-day existence. The Internet can provide considerable benefits; however, excessive use has brought about the potential for addiction and caused impairments in social, academic, financial, psychological, and work domains. Internet addiction (IA) is defined as the lack of ability to control Internet use, which causes distress, is time consuming, or results in significant social problems, occupational problems, or financial impairments [1]. Psychological disturbances like loneliness, low self-esteem, poor coping capacity, anxiety, stress, and depression are also present [2-4]. Aggressive behavior can also be related to excessive Internet use [5].

IA is not a recognized disorder in the Diagnostic and Statistical Manual of Mental Disorders (DSM-5) [6], and there is no consensus on diagnosis criteria; however, some researchers suggest features such as salience, mood modification, tolerance, withdrawal, conflict, and relapse, arguing that addictions share elements of biopsychosocial processes [7]. Other often-used diagnosis criteria based on modified criteria for pathological gambling include the following: excessive preoccupation with the Internet; the need to use the Internet for increasing periods of time; unsuccessful efforts to control Internet use; feeling restless, moody, depressed, or irritable when attempting to cut down Internet use; staying online longer than originally intended; loss of a significant relationship, job, or educational opportunity; lying to others to conceal the extent of involvement with the Internet; and using the Internet to escape from problems or to relieve a dysphoric mood. It is considered addiction when 5 or more criteria are present over a 6-month period [8,9].

Given that there are no official diagnosis criteria, researchers have validated several instruments to assess IA, and international prevalence rates vary greatly. The most-used questionnaires are the following: the Young Internet Addiction Test (IAT) [10], the Compulsive Internet Use Scale (CIUS) [11], the Excessive Internet Use Scale (EIU) [12], the Problematic Internet Use Questionnaire (PIUQ) [13], the Chen Internet Addiction Scale (CIAS) [14], The Addiction Profile Index Internet Addiction Form-Screening Version (BAPINT-SV) [15], the Internet Addiction Proneness Scale (KS scale) [16], and Young's Diagnostic Questionnaire (YDQ) [8]. Accordingly, the worldwide prevalence rates of IA differ greatly and range, approximately, from 1.0% to 18.7% [17].

Anxiety disorders share features of excessive fear and anxiety and related behavioral disturbances. These symptoms cause significant distress in social, occupational, or other areas of functioning. Panic disorder (PD) involves recurrent unexpected panic attacks that are characterized by an abrupt surge of intense fear that reaches a peak in minutes, accompanied by physical and cognitive symptoms like palpitations, sweating, chest pain, fear of losing control, fear of dying, trembling, and nausea. Generalized anxiety disorder (GAD) involves excessive anxiety and worry about daily activities that the patient finds difficult to control and is associated with being easily fatigued, irritability, muscle tension, sleep disturbance, difficulty concentrating, and restlessness [6].

People with multiple dependencies such as alcohol, cigarettes, drugs, food, and sex have a higher risk of developing IA, because they have learned to deal with anxiety and difficulties through compulsive behavior [18]. Individuals with IA usually have comorbid psychiatric disorders and this association aggravates Internet use; the relationship between IA and several psychiatric disorders is significant and has awakened academic interest. Researchers have linked IA with depression [19,20-22], attention deficit and hyperactivity [23-25], generalized anxiety disorder and social anxiety disorder [23,26-28], dysthymia [26], alcohol use disorder [29], eating disorder [30], obsessive compulsive personality disorder, borderline personality disorder and avoidant personality disorder [26], and insomnia [31]. Some researchers have suggested that IA could be a symptom of another diagnosis such as anxiety or depression and not a separate disorder [4,32], and have likened IA to impulse control disorder [2,33-35]; however, others have argued that IA should be diagnosed as a primary disorder [10,36].

These comorbidities play an important role in the treatment of IA, which should emphasize the psychiatric condition and treat abusive Internet use [19]. Studies highlight that IA causes damage in social, physical, and mental aspects of life, generating job loss, divorce, family disagreements, social isolation, academic failure, abandonment or expulsion from school [37,38], insomnia, musculoskeletal pain, tension headaches, malnutrition, fatigue, and blurred vision [31] and cognitive impairments like inattention, difficulty concentrating, procrastination, and incomplete tasks [39,40].

Treatments

Some pharmacological [41,42] and psychotherapeutic [4,18,43-46] treatments have been proposed and recommended for IA both separately and together [47]. Substantial addiction and IA can share the same neurobiological mechanism, so in this sense, addictive behavior medications can help other dependencies [3]. Medications such as escitalopram [48], citalopram [49], bupropion [41,50], olanzapine [51], quetiapine[52], naltrexone [53], methylphenidate [54], and memantine [55] have all been used to treat IA.

Cognitive-behavioral therapy (CBT) has been shown to be effective in treating IA and has been suggested in many studies [18,43,56-58]. CBT highlights the relationship between thoughts, emotions, and behaviors, and teaches patients to pay attention to these and to be ready to identify addictive behavior triggers through their thoughts and feelings. CBT psychotherapists teach coping styles, and promote adherence to treatment, changing behaviors, and preventing relapses [58]. As a treatment for IA, some researchers have suggested

traditional CBT [43,44,59-62], CBT and counseling [63], CBT with electroacupuncture (EA) [64,65], CBT and motivational interviewing (MI) [66], CBT and medication [59,61,67], cognitive or behavioral therapy [68], and a modified CBT program titled short-term treatment of Internet and computer addiction (STICA) with individual and group interventions [44].

Group psychotherapy and hospitalization for detoxification are also models of treatment for IA [5]; in addition, multimodal approaches using CBT, psychotherapy with families, treatment of comorbidities, medications, and hospitalization are also suggested [69].

Therefore, the main objective of this study is to test the efficacy of a treatment for PD or GAD and IA involving pharmacotherapy and modified CBT. A secondary aim is to produce clinical research data to corroborate the recognition of IA as a behavioral addiction and ascertain the nature of the relationship between anxiety disorders and IA.

Methods

The inclusion criteria adopted were the following: (1) patients between 18 and 65 years of age with IA; (2) a diagnosis of PD or GAD through the Mini International Psychiatric Interview (MINI), and confirmed by a psychiatrist; (3) attending and completing the initial interview; and (4) having sufficient cognitive ability to understand the instructions. Patients who did not know how to read or write, or had Axis II pathology [6], were excluded.

This study was approved by the Ethics Committee of The Federal University of Rio de Janeiro, CAAE 2704531460000526. All patients signed a consent form and attended the Laboratory of Panic and Respiration at the Institute of Psychiatry of the Federal University of Rio de Janeiro (IPUB/UFRJ).

All patients were seeking treatment for anxiety symptoms. At screening, they responded to the following scales: MINI 5.0 [70], the Hamilton Anxiety Rating Scale (HAM-A) [71], the Hamilton Depression Rating Scale (HDRS) [72], Clinical Global Impressions Scale (CGI) [73], and the Young Internet Addiction Test (IAT) [10]. IA was assessed through the IAT (scores above 50), while anxiety disorders were diagnosed by a psychiatrist. Patients were then invited to participate in this study and were forwarded for pharmacotherapy and a modified CBT protocol.

Patients were evaluated by a psychiatrist at the beginning of treatment, were permitted to take medication prescribed by the psychiatrist during treatment, and were accompanied by a psychiatrist throughout treatment.

All 39 patients underwent psychotherapy (modified CBT), which was conducted once a week for 10 weeks. The focus was to teach patients how to manage anxiety symptoms without using the Internet, and to promote conscious use of the Internet. Psychotherapy followed 4 phases: psychoeducation about anxiety and Internet use, cognitive reappraisal, behavioral modification, and prevention of relapse (Table 1).

Table 1. Description of psychotherapy.

Phase	Description
1 (3 sessions)	Psychoeducation about anxiety (PD or GAD) and Internet use, identifying triggers that increase anxiety and Internet use. Breathing retraining, breathing exercises, and strategies to manage anxiety without using the Internet. Maintenance factors: personal, situational, social, psychiatric, or occupational conditions.
2 (2 sessions)	Cognitive reappraisal of anxiety and Internet use. Daily Internet use and cognitions involving this use and anxiety. “Just a few more minutes on the Internet won’t do me any harm.” “I have to answer my friends immediately, otherwise they will not forgive me.” “If my friends don’t give “likes” on my posts or my photos, it is a signal that they don’t like me or that I did something wrong.” “If I disconnect from the Internet, I will miss important things because the best things are on the Internet.”
3 (3 sessions)	Behavioral modification, breaking routine in the use of the Internet. Training time management with a diary of Internet use, changing ways of dealing with family, friends, social activities, physical exercises, and other aspects of life. Insert positive emotion into daily activities to develop social skills to promote less Internet usage and more in-person interactions.
4 (2 sessions)	Reinforcement of continued recovery and relapse prevention through new beliefs and behaviors, social skills like assertiveness, problem solving, verbal communication, and empathy. Achievement card. Follow-up of scales.

The first phase of psychotherapy lasts 3 sessions and is focused on psychoeducation about the anxiety mechanism, identifying frightening situations, and triggers that increase anxiety and problematic Internet use. The focus is on teaching breathing retraining through breathing exercises and strategies, without using the Internet to deal with anxious thoughts and situations. During this phase, patients learn to identify and accept emotions and to stop fighting against their anxiety. Patients come to understand their anxiety and its relationship to Internet use through self-monitoring of their Internet use during situations involving anxiety. Other maintenance factors related to Internet abuse and anxiety are also explored. These factors can include personal, situational, social, psychiatric, or occupational conditions.

The second phase pertains to cognitive reappraisal of anxiety and Internet use. During this stage, patients think about their daily Internet use, cognitions involved in this use, and anxiety. Cognitive distortions are identified and the patient comes to understand that distortions such as the following contribute to their excessive use of the Internet: “Just a few more minutes on the Internet won’t do me any harm”; “I have to answer my friends immediately, otherwise they will not forgive me”; “If my friends don’t give “likes” on my posts or my photos, it is a signal that they don’t like me or that I did something wrong”;

and "If I disconnect from the Internet, I will miss important things because the best things are on the Internet." All thoughts related to anxiety and Internet use are restructured and new thoughts are proposed; alternative beliefs are generated over 2 sessions.

The third phase (3 sessions) involves behavioral modification with exposure to feared/ansiogenic situations, time management training, and proposal of a diary of Internet use. Behavioral modification involves breaking routines in the use of the Internet, and includes changing ways of dealing with family, friends, social activities, physical exercise, and other aspects of life. All components of situations are analyzed, and replaced or removed as necessary to do things differently and successfully change old ways of functioning. Another important element of this stage is the insertion of positive emotions into daily activities to develop social skills, so as to promote less Internet usage and more in-person interactions. According to positive psychology, enhancing positive emotion increases resilience, helping to reduce signs and symptoms of anxiety and depression and prevent relapse [74].

The fourth phase lasts 2 sessions with a focus on continued recovery and relapse prevention by reinforcing new beliefs and behaviors, and social skills such as assertiveness, problem solving, verbal communication, and empathy. Achievements/improvements are registered on a card (achievement card) and patients are encouraged to continue putting into practice what they have learned in psychotherapy. In the last session, volunteers responded to the same scales used at the start of treatment (IAT, HAM-A, HAM-D, and CGI), to follow up and verify improvements in scale scores. In addition to improvements in scale scores, other important criteria were reduced time spent on the Internet, increased in-person interactions, and in particular, reduced need to use the Internet to escape from problems or manage anxiety.

Results

This open trial study proposed pharmacological and psychotherapeutic interventions to treat patients diagnosed with PD or GAD and IA. Initially, 41 patients fulfilled criteria and were selected to receive psychotherapy treatment for PD or GAD and IA; however, two did not proceed with treatment (a 33-year-old male taxi driver with PD and IA who moved to another state after the third session; and a 36-year-old female with PD and IA as well as other diagnoses such as an eating disorder and recurrent depression, who attended only 2 sessions of psychotherapy). The other 39 patients attended all sessions; demographic characteristics are presented in Table 2.

Psychiatrists prescribed medications to treat PD or GAD and IA. Some of the medications used were antidepressants such as fluoxetine, sertraline, venlafaxine, desvenlafaxine, paroxetine, escitalopram, zolpidem, and duloxetine; anxiolytics such as clonazepam and alprazolam; psychostimulants such as methylphenidate; and antipsychotics such as quetiapine.

Of the 39 patients, 25 were diagnosed with PD and 14 with GAD, besides also having IA. Before treatment, anxiety levels on the HAM-A suggested severe anxiety, with an average score of 34.26 (SD 6.13); after treatment, the average score was 15.03 (SD 3.88). The IAT average score at the beginning of treatment was 67.67 (SD 7.69), indicating problematic Internet use; after the sessions, the average IAT score was 37.56 (SD 9.32), indicating moderate Internet use and a significate improvement in addiction. The mean HDRS score at baseline was 16.72 (SD 5.56), suggesting mild depression, whereas after treatment the mean score was 7.28 (SD 2.52), indicating no depression. Results of the *t* tests comparing scores before and after treatment are reported in Table 3.

Table 2. Sample characteristics.

Characteristic	Mean (SD) or n (%)
Age	28.56 (5.93) (range 19–42)
Female	27 (69%)
Male	12 (31%)
Elementary school	10 (26%)
High school	29 (74%)
Single	28 (72%)
Married	10 (26%)
Widow	1 (3%)
Student/employed	36 (92%)
Unemployed	3 (8%)

Table 3. Results of *t* -tests comparing scores before and after treatment.

	Mean (SD)		*t* -test	
	Baseline	Final	*t*	*P* value
IAT[a]	67.67 (7.69)	37.56 (9.32)	13.61	< .001
HDRS[b]	16.72 (5.56)	7.28 (2.52)	8.94	< .001
HAM-A[c]	34.26 (6.13)	15.03 (3.88)	13.62	< .001
CGI[d]	5.15 (0.65)	1.10 (0.24)	27.62	< .001

[a]IAT: Internet Addiction Test

[b]HDRS: Hamilton Depression Rating Scales

[c]HAM-A: Hamilton Anxiety Rating Scale

[d]CGI: Clinical Global Impressions Scale

Correlations between scale scores were also computed. The correlation between scores on the IAT and HAM-A was .724, between scores on the HAM-A and HDRS was .815, and between scores on the IAT and HDRS was .535.

At the end of psychotherapy, all patients felt very positive about their treatment and were very confident, after having recovered their social lives. Patients showed improvements in anxiety symptoms and managing anxiety without use of the Internet. Internet use after treatment became conscious and all patients were classified as mild users. These achievements show that patients were able to recover healthy functioning.

Discussion

In the present study, the authors described a protocol for modified CBT treatment, examined the effects of this treatment and pharmacotherapy on 39 patients with PD/GAD and IA, and analyzed the relationship between anxiety and IA. Despite controversy regarding the recognition of IA as an official disorder, the harmful effects of this behavioral addiction are highlighted in several studies [75-80]. The psychotherapy protocol was shown to be effective in the treatment of anxiety and IA, since all patients learned to manage anxiety without the Internet and showed conscious use at the end of the sessions.

Several studies have confirmed the association between depression and IA [19-23,27]; however, few studies have explored the association between anxiety and IA [26,81,82]. Imaging studies indicate that IA functions similarly to impulse control disorder; magnetic resonance imaging has shown that the areas activated when an individual with IA has the urge to use the Internet are the same areas activated by addictive substances [5]. At the same time, anxiety plays an important role in increasing Internet usage and strengthening the addiction. The authors highlighted the relationship between anxiety disorders and IA through the correlation shown (.724), which reflects the fact that beliefs and behaviors related to anxiety have an important impact on Internet use and contact with the world.

Previous treatments for IA have been described in the literature such as CBT [45,56,60,83], CBT and medication [59,67,68], and multimodal programs involving individual and group therapy, counselling, and family therapy [44,61,84].

A limitation of the study was the small sample size (39 participants); however, results showed the effectiveness of the proposed treatment, both in reducing symptoms of anxiety and promoting healthy Internet use, to improve IA in patients. Furthermore, this study is the first published research on IA treatment in a Brazilian population.

Future research should identify possible treatments for IA using new strategies and approaches, such as gestalt, counselling, family therapy, mindfulness, psychodynamic therapies, positive psychology, and transdiagnostic treatment. Investigation and analysis should also be conducted to develop new treatments for specific populations in which IA has a harmful impact, such as couples with marital problems, individuals who suffer from insomnia, individuals with attention deficit disorder, and individuals with other addictive behaviors, such as smoking, drug use, eating, sex, or shopping.

Our findings suggest that pharmacotherapy and the developed protocol of psychotherapy in the treatment of patients with anxiety and IA were effective strategies. Improvement was remarkable due to complete engagement of patients in therapy, which contributed to the success of treatment from a behavioral perspective, and gave patients confidence to continue and to manage Internet use in their lives.

IA is increasing around the world and in some countries, such as South Korea and China, it is considered a public health condition. In this sense, effective treatments should be proposed and reported that promote conscious use of the Internet, and involve valuing family, friends, a social life, and physical exercise. As such, Internet use should be conscious so as not to become abuse, and interaction over the Internet should reinforce and expand in-person interactions.

Conflicts of Interest

None declared.

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Abbreviations

BAPINT-SV: Addiction Profile Index Internet Addiction Form-Screening Version
CBT: cognitive-behavior therapy
CGI: Clinical Global Impressions Scale
CIAS: Chen Internet Addiction Scale
CIUS: Compulsive Internet Use Scale
DSM: Diagnostic and Statistical Manual of Mental Disorders
EA: electroacupuncture
EIU: Excessive Internet Use Scale
GAD: generalized anxiety disorder
HAM-A: Hamilton Anxiety Rating Scale
HDRS: Hamilton Depression Rating Scale
IA: internet addiction
IAT: Internet Addiction Test
IPUB/UFRJ: Institute of Psychiatry of the Federal University of Rio de Janeiro
MI: motivational interviewing
MINI: Mini International Psychiatric Interview
PD: panic disorder
PIUQ: Problematic Internet Use Questionnaire
STICA: short-term treatment of Internet and computer addiction
YDQ: Young's Diagnostic Questionnaire

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Original Paper

How to Conduct Multimethod Field Studies in the Operating Room: The iPad Combined With a Survey App as a Valid and Reliable Data Collection Tool

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Abstract

Background: Tablet computers such as the Apple iPad are progressively replacing traditional paper-and-pencil-based data collection. We combined the iPad with the ready-to-use survey software, iSurvey (from Harvestyourdata), to create a straightforward tool for data collection during the Anesthesia Pre-Induction Checklist (APIC) study, a hospital-wide multimethod intervention study involving observation of team performance and team member surveys in the operating room (OR).

Objective: We aimed to provide an analysis of the factors that led to the use of the iPad- and iSurvey-based tool for data collection, illustrate our experiences with the use of this data collection tool, and report the results of an expert survey about user experience with this tool.

Methods: We used an iPad- and iSurvey-based tool to observe anesthesia inductions conducted by 205 teams (N=557 team members) in the OR. In Phase 1, expert raters used the iPad- and iSurvey-based tool to rate team performance during anesthesia inductions, and anesthesia team members were asked to indicate their perceptions after the inductions. In Phase 2, we surveyed the expert raters about their perceptions regarding the use of the iPad- and iSurvey-based tool to observe, rate, and survey teams in the ORs.

Results: The results of Phase 1 showed that training data collectors on the iPad- and iSurvey-based data collection tool was effortless and there were no serious problems during data collection, upload, download, and export. Interrater agreement of the combined data collection tool was found to be very high for the team observations (median Fleiss' kappa=0.88, 95% CI 0.78-1.00). The results of the follow-up expert rater survey (Phase 2) showed that the raters did not prefer a paper-and-pencil-based data collection method they had used during other earlier studies over the iPad- and iSurvey-based tool (median response 1, IQR 1-1; 1=do not agree, 2=somewhat disagree, 3=neutral, 4=somewhat agree, 5=fully agree). They found the iPad (median 5, IQR 4.5-5) and iSurvey (median 4, IQR 4-5) to be working flawlessly and easy to use (median 5, IQR 4-5). Expert ratings also showed that the anesthesia team members (ie, the surveyed doctors and nurses) who used the iPad- and iSurvey-based tool in the OR liked it (median 4, IQR 3-4.5).

Conclusions: The combination of the iPad and iSurvey provides an efficient and unobtrusive method to observe teams in their natural environment in the OR and to survey team members immediately after completing their task (ie, anesthesia induction). The expert raters positively evaluated the use of the device and user perceptions. Considering these comprehensive results, we can recommend the use of the iPad- and iSurvey-based tool for studying team performance and team member perceptions in the OR.

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KEYWORDS

data collection; empirical research; observation; computers; informatics; anesthesiology

Introduction

The use of computer-based data collection tools has increased rapidly over the past decade, and these tools are progressively replacing paper-and-pencil-based methods [1,2].

The Apple iPad has become a revolutionary device in terms of ease of use, versatility, and availability. Useful applications in clinical practice [3-7] and medical education [8-12] have been reported. However, evidence is lacking on whether the iPad may also provide a suitable data collection tool to assess teamwork and team performance in the operating room (OR). This is problematic as a better understanding of teamwork and performance in the OR is critical for patient safety [13]. Although there are some studies that have investigated teamwork in the OR or conducted field-based experiments with health care teams [14-17], no study has outlined the specific methodological requirements that are necessary to conduct such studies. This may limit methodological knowledge and thus impede the growth of further research in this area.

We used the iPad loaded with the iSurvey app for data collection during an extensive intervention study in the OR that included data from multiple sources. Specifically, we used the combination of the iPad and iSurvey to conduct behavioral observations of teamwork and performance during systematic observations of anesthesia inductions and to assess team member perceptions regarding key teamwork aspects (eg, information exchange, knowledge of critical information, perception of safety). These assessments involved a large number of questions, which would have made a paper-and-pencil-based approach cumbersome and error-prone, considering that we collected data in various OR areas of an academic hospital [18]. Another key challenge was the fact that we aimed to investigate anesthesia teams before, during, and after anesthesia induction. This required the use of an unobtrusive and yet reliable method to assess data [19].

In this paper, we aim to provide a detailed analysis of the factors that led to the use of the of iPad and iSurvey as our combined data collection tool, illustrate our experiences with the use of this data collection tool during the intervention study, and report the results of a follow-up study conducted with the expert raters who used the tool. With the results of this research, we contribute further methodological insights on how to conduct multimethod field studies in the OR.

Methods

The following sections describe two study phases. In Phase 1, we used the iPad and iSurvey for a hospital-wide intervention study in which we evaluated a newly developed anesthesia pre-induction checklist (APIC study [18]). In Phase 2, we conducted a follow-up study to assess expert raters' perceptions and experiences of using the iPad and iSurvey as a data collection tool in the OR.

Phase 1: The APIC Study

In the APIC study [18], we tested whether the iPad loaded with iSurvey software would be a suitable tool to assess team performance and team member perceptions in the OR. Findings are based on an intervention study that tested the effectiveness of an Anesthesia Pre-Induction Checklist (APIC) using a control group design. We introduced the APIC to provide a check and briefing of safety-critical items immediately before the induction of anesthesia. The key aims of the checklist are to avoid omission errors and to improve situation awareness by promoting a shared mental model between all members of the anesthesia team.

The APIC study featured a multimethod approach comprising (1) onsite systematic observations of anesthesia inductions and (2) surveys of the observed anesthesia team members conducted immediately after the onsite observations.

We compared data from teams who used the APIC (intervention group) during anesthesia induction with teams who did not use the APIC (control group). Specifically, we tested the effects of the APIC on communication and technical performance of anesthesia teams and team members' awareness of critical information, perceptions of safety, and perceptions of teamwork. Ethics approval was given by the ethics committee of the Canton of Zurich (KEK StV-Nr. 07/12), Zurich, Switzerland.

Participants

We observed a total of 205 anesthesia inductions in seven OR areas at the University Hospital Zurich, Zurich, Switzerland. We observed 105 teams (including a total of 285 team members, ie, doctors and nurses) before, and 100 teams (272 team members) after the introduction of the APIC.

Procedure and Measures

In the following section, we will outline (1) factors that led to the decision to use an electronic data collection tool, (2) requirements that our desired data collection tool needed to fulfill, (3) factors that led to the use of the iSurvey software specifically, (4) how we created the iPad- and iSurvey-based data collection tool, and (5) how we applied the iPad- and iSurvey-based tool during the APIC study.

Reasons to Use an Electronic Data Collection Tool

The decision to use an electronic data collection tool was based on the following considerations. First, we planned to observe anesthesia inductions in seven different operating areas situated in multiple locations of an academic hospital. Second, the study required large numbers of observations and involved an extensive data collection protocol (more than 60 items per observation). Third, the anesthesia teams were observed during and surveyed immediately after the anesthesia induction, which required us to use a fast and unobtrusive way to assess data.

A paper-and-pencil–based data collection method would have required the multiple data collectors to handle and keep track of large amounts of paper. We reasoned that this would have made data collection, storage, and management more time and

energy consuming and more prone to errors when compared to an electronic data collection method. We thus sought a simple and reliable electronic method to collect our data.

Requirements for the Desired Survey Software

Before deciding on a specific survey app to be used during our research project, we defined some criteria that we considered important. As wireless Internet access could not be guaranteed in all positions inside the operating areas, we required an app that provided offline data collection. Moreover, the creation of surveys had to be easy and straightforward, without the requirement of software-programming skills. The software had to be ready for data entry within a couple of seconds, and if the data collection was interrupted during an observation, the survey had to restart at the same position after pushing the start button. We also needed to be able to use a branching logic—mandatory questions that inhibit the continuation of the survey until a question has been answered and group questions to avoid switching between survey screens in order to minimize cognitive effort of the data collectors. Also, the answers had to remain saved when going back and forth between survey screens. Finally, the app also had to have a reasonable price to fit our research budget.

Reasons to Use iSurvey Software

While planning the study in February 2012, before selecting a survey app, we downloaded and evaluated all survey apps that offered a free initial download on the US and Swiss Apple App Stores, using the search terms “survey” and “data collection,” in order to identify the app that best met our previously defined requirements. We evaluated the following apps: SurveyPocket by Jeremy Przasnyski (surveyanalytics site), Polldaddy by Auttomatic, Inc. (polldaddy site), iFormbuilder by Zerion Software, Inc. (iformbuilder site), and iSurvey (Harvestyourdata, Wellington, New Zealand). We also evaluated SurveyMonkey and Qualtrics, but these providers did not offer a solution that worked offline on an iPad. We decided to use iSurvey because it was the only one of these apps that saved answers when going back and forth between screens and allowed grouping of multiple answers on a single screen.

Creation of the Data Collection Tool

Once we made the decision to use iSurvey as the software for our data collection tool, we created the survey containing all the data collection protocol questions for the study in the password-protected user area of iSurvey site. The exact number of questions asked per observation varied because we used the iSurvey function to choose a branching logic. Using this feature, the survey directs the user to a prespecified question or information screen depending on how a question is answered. Also, control questions were included to verify that an observation was within the predefined study inclusion criteria. For example, if the question “Is this an emergency situation?” was answered with “yes,” the survey was terminated because only anesthesia inductions for elective surgery, and not emergency procedures, were to be included. We also used the function of iSurvey to randomize the order of the answers to a question to minimize common survey response biases such as the tendency to respond in the same direction on a series of questions regardless of the content.

Application of the Data Collection Tool During the APIC Study

We recruited 5 attending anesthesiologists (ie, each with more than 5 years of clinical anesthesia experience) to serve as expert raters of team performance and as data collectors for the survey of team member perceptions. Prior to the observations in the ORs, we conducted a training session. This session served to (1) explain the study procedure, (2) familiarize the expert raters with the data collection tool, (3) train the raters in observational skills, and (4) test the interrater reliability of the data collection tool. We explained to the expert raters how to start the iPad and iSurvey and how to upload data after an observation. We also conducted a rating of a videotaped anesthesia induction scenario together with the expert raters. To assess interrater agreement, we recorded three multi-angle videos of anesthesia induction scenarios showing different levels of team performance in a full-scale anesthesia simulator. All 5 expert raters then independently watched and rated the videos using the data collection tool, and Fleiss’ kappa was calculated.

During the data collection phase of the study, we conducted three meetings with all expert raters to address questions pertaining to the iPad- and iSurvey-based data collection tool. During the study, the expert raters answered general questions and questions about team performance (ie, communication and clinical performance). For example, a general question was “Which team member read the checklist? Consultant/resident/nurse.” An example of a team performance question was “Did the team talk about the patient allergies? Yes/no.”

The expert raters completed nominal scale-level questions (yes/no, and different choices, multiple-choice, and single best answer; for example (different choices single best answer), “Name of the OR area the observation is taking place in. OR area #1, OR area #2, etc”.

After the observation, the expert raters handed the iPad to each observed team member, who then individually and privately answered a short survey. The individual team members answered general questions and questions about their perceptions during the induction. For example, a general question was “My anesthesia experience in years? >1, 1-5, 5-10, >10 years,” or a question about team member perceptions was “How safe did I feel during this induction?” answered on a continuous Likert-type rating scale from 0% (very unsafe) to 100% (very safe). The anesthesia team members completed nominal scale-level questions, different choices (multiple-choice and single best answer), and interval scale-level questions (continuous rating scales).

The observation and team member survey procedure for anesthesia teams in both the APIC group and the control group did not differ, and teams in both groups were observed and surveyed equally by the same expert raters. These expert raters did not participate in the anesthesia inductions they observed or in the team member surveys after the inductions. Their sole purpose was to rate the anesthesia induction and administer the team member survey to the observed team members after the induction.

The collected data were downloaded from the password-protected user area from the iSurveysoft site as a MS Excel readable CSV (comma separated value) file for analysis.

Figure 1 shows screenshots of the data collection tool used in this study. Figure 2 shows an example of how the data collection was conducted in the ORs.

Figure 1. Screenshots of the iPad- and iSurvey-based data collection tool. The left shows the tool asking the data collector to name the operating area in which the observation is taking place. The right shows example questions asked during the team member survey.

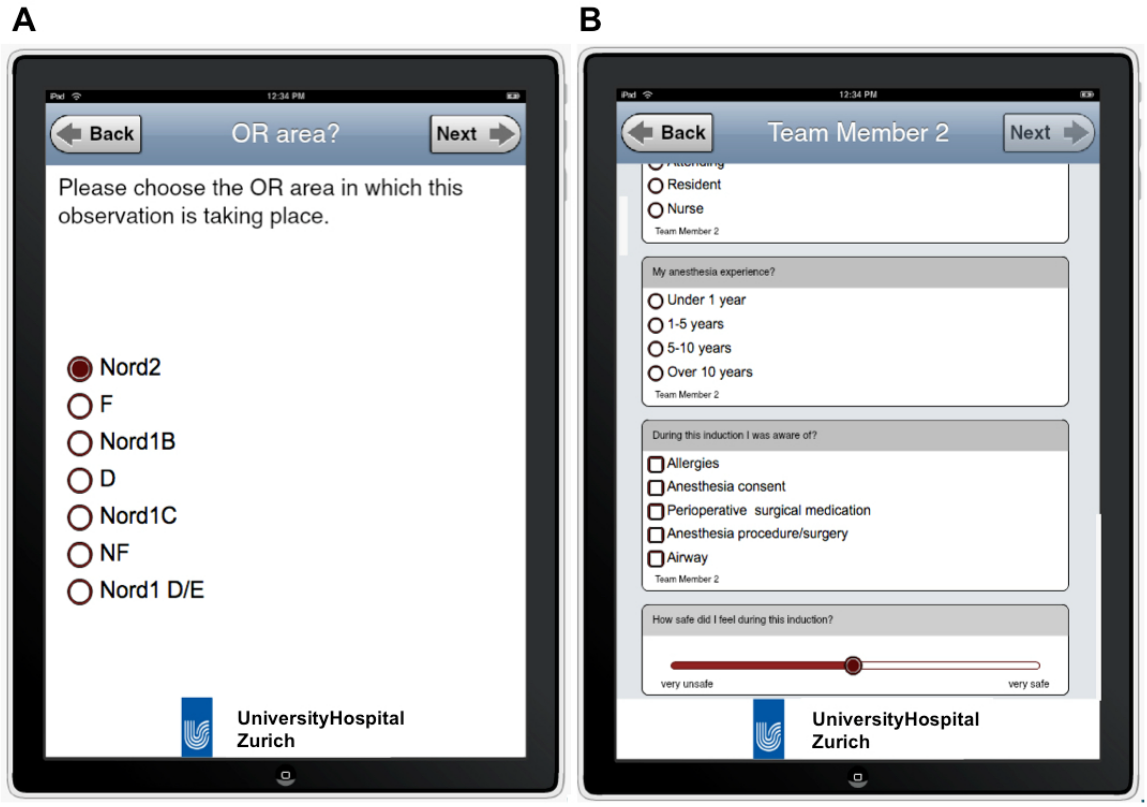

Figure 2. A data collector using the iPad- and iSurvey-based data collection tool to rate anesthesia team performance during a systematic onsite observation of team performance.

Phase 2: Expert Rater Survey

After the completion of the APIC study, we conducted a follow-up survey of the data collectors to assess their perceptions about the iPad- and iSurvey-based data collection tool. We aimed to receive a more in-depth understanding of the use of the iPad- and iSurvey-based tool and to assess the expert raters' impressions of the use of the tool.

Participants

We conducted a survey with the same 5 expert raters who used the iPad- and iSurvey-based tool in the ORs during the observations and team member surveys of the APIC study.

Procedures and Measures

An email containing a single-page MS Word document as an attachment was sent to the 5 expert raters. The survey contained a total of 7 questions: 6 questions with Likert-type rating options (1=do not agree, 2=somewhat disagree, 3=neutral, 4=somewhat agree, 5=fully agree) and one open-ended question, which enabled the expert raters to provide any kind of feedback. Table 1 contains all questions (1-7) as well as the raters' responses to each question.

Results

APIC Study (Phase 1)

During the training session conducted prior to the beginning of the data collection, 4 of the 5 data collectors were already familiar with the iPad and therefore needed to be instructed only on how to use the iSurvey app. We found that all expert raters, including 1 rater who was not previously familiar with the iPad, were easily and quickly familiarized and immediately able to use the iPad- and iSurvey-based data collection tool. Interrater agreement of the data collection tool was found to be very high for each of the three videotaped scenarios. Median Fleiss' kappa for the three scenarios was 0.88 (95% CI, 0.78-1.00).

During the three meetings we conducted with the expert raters during the study, they did not report any problems and the data collection was completed without any problems. The expert raters and team members were able to answer the more than 60 questions of the data collection protocol in an average time of 25 minutes. The download of the data was convenient and fast, without any interruptions. We were able to export the data into MS Excel (Mac 2011).

For our 131 days of data collection, we needed to subscribe to iSurvey for 5 months. iSurvey is available from US $89 per month (December 2015). One subscription allows one survey to be completed up to 3000 times per month on an unlimited number of devices. Also, we acquired one iPad for each expert rater. The iPad is available from US $269 (iPad mini 2 WiFi, December 2015).

Expert Survey (Phase 2)

The results of the expert survey contained Likert-type ratings and open-ended responses concerning multiple data collection aspects. The majority of raters were positive that the anesthesia team members (ie, the surveyed doctors and nurses) using the iPad- and iSurvey-based tool in the ORs liked it (see Table 1). Most clearly, the raters preferred the iPad- and iSurvey-based tool over a paper-and-pencil–based method they had used during other, earlier studies (median response 1, interquartile range [IQR] 1-1). Furthermore, they found the iPad (median 5, IQR 4.5-5) and iSurvey (median 4, IQR 4-5) worked flawlessly and was easy to use (median 5, IQR 4-5). Expert ratings also showed that the anesthesia team members (ie, the surveyed doctors and nurses) who used the iPad- and iSurvey-based tool in the ORs liked it (median 4, IQR 3-4.5). In addition, all observers provided answers to the open-ended question (question 7). Their unedited answers are given in Table 2.

Table 1. Survey of raters' experience using iSurvey software on iPad tablet computers[a].

Question	Rater 1	Rater 2	Rater 3	Rater 4	Rater 5	Median (IQR)
1. I found the iPad loaded with iSurvey easy to use for observing teams in the operating room.	5	5	5	4	4	5 (4-5)
2. The app iSurvey worked flawlessly during the study.	5	4	5	5	4	5 (4-5)
3. The iPad worked flawlessly during the study.	5	4	5	5	5	5 (4.5-5)
4. I would prefer to use paper and pencil over an iPad- and iSurvey-based tool when observing teams in the operating rooms.	1	1	1	1	1	1 (1-1)
5. The anesthesia team members liked using the iPad- and iSurvey-based tool to answer the questions in the operating rooms.	4	3	5	3	4	4 (3-4.5)
6. The anesthesia team members answering the questions had problems using the iPad- and iSurvey-based tool.	1	4	1	1	2	1 (1-3)
7. Were there any problems using the iPad in the operating rooms? Do you have any other remarks?	Open-ended question					

[a]1=do not agree, 2=somewhat disagree, 3=neutral, 4=somewhat agree, 5=fully agree.

Table 2. Unedited answers of the 5 observers to the open-ended question "Were there any problems using the iPad in the operating rooms? Do you have any other remarks?"

Number of observers	Unedited answer	Summary
1	No problems observed during the use of the iPad-based survey. Data can be better anonymized when using an iPad based survey, because as soon as the team members completed their survey the answers of the participant were not visible to us? (and did not have to be stored somewhere in the OR).	No problems; Data safety advantages
2	No real problems, though some hesitation by the team members could be noted, especially by means of hygiene and removal of gloves before use.	No serious problems; Uncertainties about hygiene
3	No problems, no remarks	No problems
4	During a certain period of time, the uploading didn't work the way it should have, but in the end, everything was fine!	Problems with data upload
5	Having a tool of the size of a regular iPad is not always helpful in the busy routine of the OR (where to put it when finishing the survey…)	Remark about the size of the data collection tool

Discussion

Principal Findings

We conducted two studies that showed that the combination of iPad and iSurvey provided an efficient and effective way to collect observational and survey data in the OR. Based on the results of an intervention study that evaluated the use of an Anesthesia Pre-Induction Checklist (APIC), we found that the tool was suitable for both systematic observations of team performance during anesthesia inductions as well as team member surveys thereafter. Based on the results of a follow-up survey, we found that the iPad- and iSurvey-based tool was well accepted and easily used by the expert observers.

Phase 1: Using the iPad and iSurvey to Observe Team Performance and Assess Team Member Perceptions

Comparing the results of the individual expert raters' ratings of videotaped pre-induction scenarios allowed us to assess interrater reliability of the iPad- and iSurvey-based data collection tool. The high interrater reliability scores showed that (1) immediately after the rater training, expert raters were able to use the combined tool in the OR, and (2) the obtained data provided a reliable assessment of team performance during anesthesia inductions in the OR.

The training of the data collectors was effortless with the iPad- and iSurvey-based tool being immediately intuitive to the expert raters. The multi-touch finger-sensitive interface of the iPad is used in many Apple devices (eg, iPod, iPhone, Apple Watch), and many people are familiar with the use of this interface [20-22].

iSurvey integrates the name of the device from which data were entered in each rating record, for example, iPad #1. This made it easy to create subgroup analyses, for example, to compare the results of individual observers and to compute interrater agreement.

Phase 2: Experiences of Expert Raters

The results of the expert rater survey showed that the majority of raters either completely or somewhat agreed that both the iPad hardware and iSurvey software worked flawlessly and they found it easy to use for observing teams in the OR. The data also revealed some additional experiences on the part of raters. For example, one rater stated that data collection is safer compared to a paper-and-pencil-based method, because unlike on paper, after the data has been entered it is not present on paper sheets lying around in the OR, where they could possibly be read by another person. Some minor problems were reported as well that are noteworthy to discuss. First, one rater stated that some study participants were hesitant to use the tool during the induction and remove their gloves before using the iPad. This is an important factor that comes into play when team members have to complete surveys immediately after an anesthesia induction. To facilitate ease-of-use in sterile environments, it might be useful to use a technique for sterile iPad use [5].

Another rater commented that the upload did not work during a certain time, which may have been caused by a temporary unavailability of iSurvey due to maintenance. This had been communicated to us in advance by Harvestyourdata and lasted for only a couple of hours. All data collected during this timeframe were saved on the iPads and could be uploaded once the service became available again.

Finally, 1 rater pointed out that there were some problems with bringing the iPad to the ORs and not knowing where to put it after an observation. This problem could be mitigated by using iSurvey on an iPhone, iPad mini, or iPod in future research projects. These devices are smaller in size but just as powerful.

In conclusion, this survey showed that the iPad- and iSurvey-based tool was well accepted and considered to work well by the sharp-end expert observers, who used the tool during an actual study. There were no major problems and the impressions of the expert raters were that the surveyed team members had neutral or positive sentiments towards the iPad- and iSurvey-based tool, but no negative sentiments.

Theoretical and Practical Implications

The combined use of iPad and iSurvey enabled us to collect data seamlessly, which was especially important in the OR, which is an environment that allows only limited disturbance by researchers. Furthermore, the tool eliminated the need to transcribe data from paper sheets to electronic data files. The use of an electronic iPad- and iSurvey-based data collection tool may provide some important advantages over a

pencil-and-paper–based approach. First, rater accuracy may be improved because there are no paper-based data collection forms to keep track of. Second, once the observations are completed, the dataset can be downloaded as a ready-to-use data file thus avoiding the risk of data transcription errors or errors in the assignment of papersheets to the correct dataset (likely to occur when paper-based observations need to be copied into electronic data files). Third, the number of datasets, which can be handled electronically, is practically unlimited. Furthermore, additional data points such as GPS position can be gathered by an electronic tool that might serve as a cross-check to the rater responses and thus further improve accuracy.

Our findings contribute to a better understanding of how multimethod field studies can be conducted and implemented in a context as sensitive as the OR. Prior research has focused on what kind of teamwork patterns should be observed in order to assess team performance in the OR [23], but few studies have outlined how to conduct such research. Our findings show that when teamwork episodes consist of short fragmented cycles (ie, during and after an anesthesia induction) and are distributed across multiple locations, a tablet computer such as the iPad combined with a ready-to-use survey app (iSurvey) is well suited to assess team performance and individual teamwork perceptions. We thus conclude that using the iPad combined with iSurvey may not only be a useful tool to assess data in the OR and other health care settings but in many other high-risk and action team settings such as aviation, mining, or the military. Given that our observers were also experienced clinicians who had to monitor the performance of the observed anesthesia team, the use of an iPad provided an effortless and very convenient way to carry and store the data collection tool at any time during the data assessment.

We also found that combining iPad and iSurvey can be used to observe as well as to survey study participants. Thus, using the iPad as a data collection tool can help triangulate different data sources to more accurately capture teamwork processes in the OR [24].

Limitations

Notably, our study has some limitations, which we will outline below. First, it must be noted that we evaluated the use of the iPad- and iSurvey-based tool using parsimonious rating scales to assess team performance and team member perceptions in the OR. In order to use more sophisticated observation methods such as time-based or event-based rating methods (ie, counting and logging the frequency of a certain behavior occurring during a specified time frame), time-logging of a variety of behavioral markers during a team interaction period is necessary. For example, observation studies in the OR looking at team coordination or communication [25] have used coding schemes consisting of 12 or more behavioral codes. Using more complex coding schemes may surpass the capabilities of a ready-to-use survey software and require the use of a behavioral coding software. Future research may address how more complex behavioral coding manuals can be used in combination with tablet computers to facilitate real-time coding in the OR.

The results of this work are based on a single center study, and the expert rater survey featured a small sample size. Future work studying end user perceptions about electronic data collection tools should feature a larger sample size.

Like any data collection method, the use of the iPad- and iSurvey-based data collection tool must be evaluated and approved by the responsible ethics committee before the beginning of a research project. The use of an iPad- and iSurvey-based data collection tool, depending on the regulation in place, may not be possible in all countries for all research projects. One safety-critical limitation of an iPad- and iSurvey-based data collection tool is its use in an environment near a strong magnetic field, for example, a magnetic resonance imaging device. Electrical devices exhibit substantial magnetic field interactions such as translational attraction and heating. This could potentially injure a patient or a caregiver.

Further regulations that govern the use of computerized data collection protocols should be taken into account before conducting a study that involves the use of iPads in a hospital context. Consulting the Food and Drug Administration Code of Federal Regulations title 21 [26] and the European Medicines Agency guidelines for Good Clinical Practice may be important [27]. In all cases it should be ascertained that collecting data with iPad and iSurvey is in accordance with standards for human subject research as derived from the World Medical Association Declaration of Helsinki - Ethical Principles for Medical Research Involving Human Subjects [28].

Conclusion

Based on the results of a large field study conducted in multiple operating room areas of a teaching hospital and a follow-up study of the expert raters who used the tool, we outlined and evaluated the combined use of iPad and iSurvey. We found that the use of the iPad- and iSurvey-based tool was suitable to observe teams in their natural environment, to collect clinical performance and communication data, and to survey team members immediately after they completed their task (ie, anesthesia induction). Additionally, it was positively evaluated by expert raters. Considering these comprehensive results, we can recommend the use of the iPad- and iSurvey-based tool for studying team performance and team member perceptions in the OR.

Conflicts of Interest

None declared.

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Abbreviations

APIC: Anesthesia Pre-Induction Checklist
CFR: Code of Federal Regulations
IQR: interquartile range
KEK: Kantonale Ethik Kommission (Cantonal Ethics Commission)
OR: operating room

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Original Paper

An Electronic Wellness Program to Improve Diet and Exercise in College Students: A Pilot Study

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Abstract

Background: In transitioning from adolescence to adulthood, college students are faced with significant challenges to their health habits. Independence, stress, and perceived lack of time by college students have been known to result in poor eating and exercise habits, which can lead to increased disease risk.

Objective: To assess the feasibility and to determine preliminary efficacy of an electronic wellness program in improving diet and physical activity in college students.

Methods: A 24-week diet and physical activity program was delivered via email to 148 college students. The intervention involved weekly, tailored, and interactive diet and physical activity goals. The control group received nondiet and nonexercise-related health fact sheets. Anthropometric and blood pressure measurements, as well as food frequency and physical activity surveys were conducted at baseline, week 12, and week 24. Students' choice of fruit as a snack was also monitored at study visits.

Results: Students were 18-20 years old, 69% female, and from a diverse college campus (46% Caucasian, 23% Asian, 20% African American, 11% other). At week 24, 84% of students reported reading at least half of all emails. Mean change (standard error [SE]) from baseline of saturated fat intake was marginally significant between the treatment groups at week 24, 0.7 (SE 0.42) % kcal for control and -0.3 (SE 0.30) % kcal for intervention (P=0.048). A significant difference in percent of snacks chosen that were fruit (χ^2_1, N=221 = 11.7, P<0.001) was detected between the intervention and control group at week 24.

Conclusions: Use of an electronic wellness program is feasible in college students and resulted in a decrease in saturated fat intake and an increase in observed fruit intake compared to a control group.

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KEYWORDS

college students, eHealth, telemedicine, diet, intervention studies, obesity, exercise

Introduction

The phrase "freshman 15" was coined to describe weight gain associated with poor health habits in US college students. Studies of this phenomenon have identified an average of a 1-4 kg weight gain during the first year of college [1-4]. In the United States, this can result in increased proportions of students being classified as overweight, from 21% at the beginning of freshman year to 32% at the end of the first school year [1].

This weight gain can lead to obesity in adulthood. Specifically, the risk of being obese as an adult is 4 times as high for an overweight or obese adolescent, compared to a normal-weight adolescent [5]. Obese adolescents are at increased risk for disease as an adult, particularly cardiovascular disease [6-8].

College is a time of increasing independence and growth. During this period, students may make poor diet choices such as skipping breakfast, consuming salty and sugary snacks, and increasing alcohol consumption, and may fail to meet physical activity guidelines [1,4,9-13]. According to 2013 data from the National College Health Assessment, only 6% of college students report eating at least 5 servings of fruits and vegetables daily and less than half meet recommendations for exercise [14]. Such health behaviors result in a generation entering adulthood at risk for obesity, cardiovascular disease, and cancer. Clearly, there is a need to improve diet and exercise habits of college students.

Contemporary college students are a technologically wired generation having been born and raised in the age of home computers and portable electronic devices. Electronic health interventions (eHealth) have the advantage of reaching larger numbers of individuals with fewer resources than face-to-face interventions [15]. The duration of eHealth studies in college students has ranged from 30 days [16] to 2 years [17], with most studies reporting a duration of 12 weeks [18-22]. A variety of eHealth methods including virtual technology, graded classroom assignments, and Web-based coaching using Facebook, email, and SMS text messaging, have been successful in significantly increasing physical activity in college students [18,19,22,23]. SMS text messaging requires short messages and, therefore, education and graphics are limited. Facebook has also been used to encourage weight loss in college students [17,24]. Facebook allows for graphics and social networking but control of privacy may be an issue for users. Weight loss programs employing other Internet technology have been used successfully in overweight and obese college students [20,21]. Previous attempts to prevent excess weight gain by college students using eHealth have had mixed results [13,25-28]. Green [26] and Kattelmann [28] were able to demonstrate improvements in diet and physical activity behaviors, but no difference in weight gain or body measures, while Gow [25] and Levitsky [27] demonstrated the prevention of increases in body mass.

The objective of this pilot study was to determine to what extent a 24-week email-delivered health intervention program, which was designed to promote healthy diet and physical activity habits in college students, would be received by a diverse sample of college students and to report preliminary efficacy for diet, physical activity, and body measures. In terms of feasibility, we hypothesized that the enrollment goal (135 college students) would be met within the first 6 weeks of the semester and that > 60% of students enrolled would report reading at least 50% of the emails. Moreover, it was expected that after 24 weeks, those who received the eHealth intervention would consume more fruits and vegetables, less sugar and fat, and would report more physical activity than those in the control group. Furthermore, changes in diet and physical activity were anticipated to result in attenuation of body composition measures and improved levels of fitness.

Methods

Participants

Approval was obtained through the University Institutional Review Board (IRB). Eligible students were recruited from a large eastern university campus utilizing IRB-approved announcements via campus list-serve technology. Interested students contacted the study team for an initial phone screening to determine eligibility. Eligible students were then scheduled for an in-person informed consent visit. Students were eligible if they were enrolled in the university during the semester the study was conducted, were 18-20 years old, and had access to email. Informed consent was conducted by trained research staff at the first study visit, prior to any study procedures. Students were excluded if they were pregnant, lactating, reported a history of an eating disorder or bariatric surgery, were currently following a diet treatment plan for weight loss, or were participating in a research study that affected health behaviors. Eligible participants were randomized 2:1 and stratified by body mass index (BMI) and gender to the intervention or control group. This ratio ensured that dropouts did not affect the ability to demonstrate the feasibility of this pilot program [29].

Numbering of study weeks was linked to the baseline visit when the individual participant was randomized and began the electronic intervention, rather than on the academic calendar. Study enrollment was staggered over the first 6 weeks of the fall semester. The schedule for subsequent study visits was staggered similarly, to coincide with the follow-up surveys included in the intervention program. Participants were given a $25 gift card to a local department store for completing baseline, week 12, and week 24 study visits. Students who completed all visits were entered into a raffle for a $250 gift certificate.

Anthropometric Measures

Trained research assistants conducted bioelectrical impedance analysis, anthropometric, and blood pressure measurements. All anthropometric data were performed in triplicate on the right side. Students were asked to refrain from exercise for 2 h, and maintain a 4-h fast (2 h for water, 12 h for caffeine and alcohol) prior to anthropometric measurements. A digital scale (Scale-Tronix, serial number 5002-27460) was used to weigh participants in underclothes to the nearest 0.1 kg. A wooden portable stadiometer (Shorr Productions) was used to measure participants' height to the nearest millimeter, with the posterior heel, buttocks, shoulder blades, and head touching the vertical board of the stadiometer with the head in the Frankfurt plane [30]. The accuracy and precision of the scale and stadiometer were verified daily using two 20 kg weights and a 160 cm rod, respectively. A Gulick II fiberglass tape measure (Country Technology, model 67020) was used to measure body circumference to the nearest millimeter. Mid-neck circumference (NC) was measured between mid-cervical spine and mid-anterior neck just below the laryngeal prominence with the head in the Frankfurt plane [31]. Waist was measured at the right superior iliac crest [30]. Hip circumference was measured at the widest area across the buttocks [32]. Waist-to-hip ratio (WHR) was

calculated by dividing the waist circumference (WC) measure in cm by the hip circumference measure in cm.

In preparation for bioelectrical impedance analysis (BIA), participants were questioned about fasting status, and were asked to empty their bladder. Urine samples were collected for pregnancy tests. All external metal from the right side of the body and any personal electronic devices were removed, to avoid interference in the flow of the electrical charge from the BIA. Pregnancy, history of seizures, and heart arrhythmias were considered exclusion criteria for BIA. Two participants were deferred, one for history of arrhythmias and a second for history of seizure. A third student failed to meet fasting conditions and did not complete baseline BIA. Participants rested supine avoiding skin-to-skin contact during the entire procedure. BIA was conducted according to the manufacturer's instructions using an ImpediMed DF50 bioelectrical impedance analyzer. Briefly, skin of the right hand, wrist, foot, and ankle were scrubbed with alcohol wipes prior to placing 4 electrodes (1) midway between the distal ulna and the dorsal radius of the wrist; (2) 5 cm from the wrist electrode, toward the fingers; 3) midway between the distal tibia and dorsal fibula; and 4) 5 cm from the ankle electrode, toward the toes. Participant height, weight, sex, and age were entered into the software before conducting the BIA. Estimates of body composition were obtained from the BIA software.

Diet and Physical Fitness

Students completed demographic, psychosocial, diet, and physical activity surveys online. The diet survey consisted of a food frequency questionnaire adapted from the Block Food Frequency Questionnaire [33]. The physical activity questionnaire was adapted from the Cross-Cultural Activity Patterns Questionnaire (CCAPQ) [34]. Reliability and validity have been established for both [34,35]. Exercise was defined as moderate to vigorous physical activity as categorized in the CCAPQ [36].

To prevent adverse reactions from fasting, each participant was offered water and snacks prior to the Queen's College Step Test [37]. Snacks included fruit (6 types), cookies (4 types), and crackers and chips (11 types). Consumption of the snack was used to evaluate whether the eHealth messages resulted in actual diet change. Snacks were measured before serving to participants and again after the study visit. Postweight was subtracted from preweight to obtain total intake. Because consumption of snack was optional, frequency of consumption was used (the number of times students ate >50% of a snack divided by total snacks chosen at that visit) for each treatment group.

Seated blood pressure was measured under resting conditions using a 10 series automated blood pressure monitor (Omeron Healthcare) per the manufacturer's instructions.

The Queen's College Step Test procedure required participants to step up and down on a 41.3 cm wooden step for 3 min at 24 steps/min for males and 22 steps/min for females [37] while wearing an Omron HR-100C heart rate monitor transmitter. Step cadence was set using a metronome (Korg, model MA-30) and monitored using a Survivor III stopwatch (Accusplit, model S3MAGXLBK). Maximum oxygen utilization (VO_{2max}) in mL/kg/min, was estimated for men (VO_{2max}= 111.33 − 0.42 × heart rate in bpm) and women (VO_{2max}= 65.81 − 0.1847 × heart rate in bpm) [37]. Two intervention group members were deferred from the Queen's College Step Test due to uncontrolled asthma and injury.

Intervention

The eHealth intervention consisted of A Lifestyle Intervention via Email (ALIVE), an evidence-based Web-based behavior change program created and managed by NutritionQuest [38] and modified by the authors for use with college students. Modifications primarily eliminated or replaced work and family-oriented language. For example, references to work and family were rewritten to include campus or apartment living and dining. Social and cognitive principles inherent to ALIVE included goal setting, a focus on individual choice, direct information and goal relevance for each learner, overcoming barriers, specific action-based advice such as establishing walking groups, salience of cues, building on prior learning, repetition of core messages, and repeated practice of new behaviors to transform them into sustained habits. There was an emphasis on small, achievable, and cumulative goals, for which accomplishment builds the participant's self-efficacy to make changes and can enhance long-term maintenance.

After completing diet, physical activity, medical history, and stages of change surveys, participants received feedback comparing their responses to recommended levels of (1) fat and sugar intake, (2) fruit and vegetable intake, and (3) physical activity. As part of the ALIVE program, all participants were encouraged to select a goal related to the 3 feedback topics. Once randomized, participants in the intervention group chose one of these topics as the focus for weekly messages offering tailored small-step goals, tips for overcoming barriers to goals, health information, and social support. Web links in the intervention email also led students to their personal account on the ALIVE website, where educational information and feedback on progress were offered. Follow-up diet and physical activity surveys were delivered to participants every 12 weeks as part of ALIVE. A 24-week duration was chosen to best coincide with the school year. The intervention group received the adapted ALIVE program via weekly emails. The control group received weekly information related to nondiet, nonexercise health topics, such as distracted driving, sleep hygiene, and smoking cessation.

Data Collection

Study data were collected and managed using Research Electronic Data Capture (REDCap) hosted at Children's National Medical Center. REDCap is a secure, Web-based application that provides (1) an interface for validated data entry, (2) audit trails for tracking data manipulation and export procedures, (3) automated export procedures for data downloads to common statistical packages, and (4) procedures for importing data from external sources [39].

Data Analysis

Demographic and baseline characteristics of students in the intervention group were compared to those in the control group using *t* test for continuous variables and chi-square for

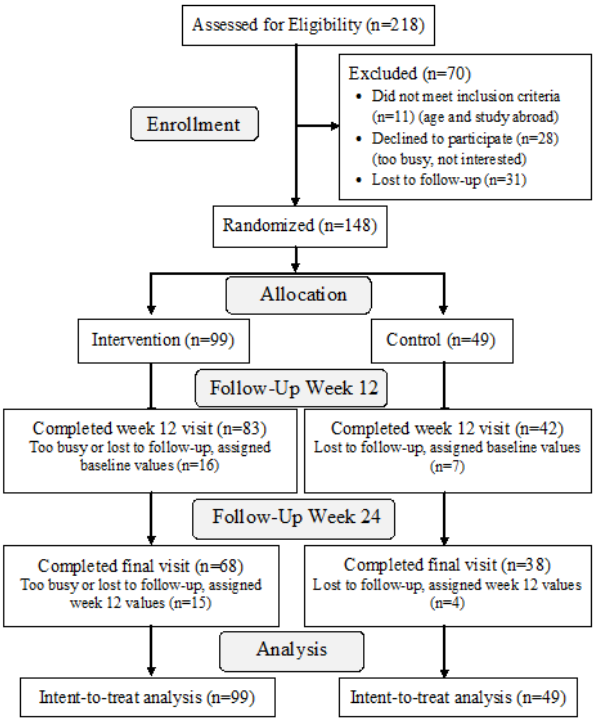
categorical variables. Chi-square was also used to determine differences in snack choice categories between the intervention and control groups. Changes over time were analyzed by repeated measures analysis of variance using a 2 (treatment: intervention, control) × 3 (time: baseline, week 12, week 24) factorial treatment design and sex and baseline BMI were controlled as covariates. Goodness-of-fit statistics were used to select the repeated measures structure that best fit the data. Significance was set at $P<.05$. The primary analysis was an intent-to-treat approach (ITT) with the last observation carried forward to fill in missing data. Secondary analyses were performed on a subgroup of completers. These participants were compliant with inclusion/exclusion criteria throughout the study period and completed all study visits and questionnaires.

Results

Participants

An IRB-approved recruitment email was sent to 10,370 students, 693 of whom responded. The recruitment goal for this pilot study was 135 to retain 100 students with an expected dropout rate of 25%. The study team was able to contact 317 students by the last enrollment day; 99 did not respond and 218 were screened for eligibility, resulting in 99 students randomized to the intervention group and 49 to the control group (Figure 1). Mean age (standard error [SE]) was 19.7 (SE 0.06) years. Study participants were primarily female and racially diverse (Table 1). The participants were generally healthy, reporting low rates (1-5%) of hypertension, hypercholesterolemia, gastrointestinal disease, and anemia. The highest prevalence of disease was asthma (17%). Dietary supplement use was reported by 31% of students. Less than 5% of students were smokers. Other than previous research experience, baseline characteristics did not differ between treatment groups (Table 1).

Figure 1. Consort flow diagram for college students recruited and retained.



Students who dropped out (intervention group: n=31, control group: n=11) had significantly higher mean BMI (24.2 [SE 0.62] kg/m^2, P=.008), WC (85.3 [SE 1.55] cm, P=.002), WHR (0.86 [SE 0.0067] cm, P=.004), and NC (34.6 [SE 0.50] cm, P=0.006), yet lower energy intake (19.8 [SE 1.31] kcal/kg, P=.011), than those who completed all visits (BMI: 22.4 [SE 0.32] kg/m^2, WC: 79.7 [SE 0.79] cm, WHR: 0.84 [SE 0.0043] cm, NC: 33.0 [SE 0.29] cm, and energy intake: 24.0 [SE 1.04] kcal/kg).

Table 1. Baseline characteristics of college students by treatment group

Characteristic		Intervention n (%)[a]	Control n (%)[a]	*P* [b]
Age (years): mean (SE[c])		19.8 (0.07)	19.6 (0.1)	.15
Sex				.93
	Female	68 (69)	33 (67)	
	Male	31 (31)	16 (33)	
Race/ethnicity				.64
	White, non-Hispanic	47 (47)	21 (43)	
	Asian	23 (23)	11 (22)	
	African-American	16 (16)	13 (26)	
	Hispanic	7 (7)	1 (2)	
	Mixed	4 (4)	2 (4)	
	Other	2 (2)	1 (2)	
School year				.15
	Freshman	3 (3)	5 (10)	
	Sophomore	38 (39)	23 (48)	
	Junior	53 (55)	19 (40)	
	Senior	3 (3)	1 (2)	
Transportation to campus				.55
	Live on campus	45 (46)	28 (57)	
	Walk/run	28 (29)	11 (22)	
	Drive	17 (17)	8 (16)	
	Bike	8 (8)	2 (4)	
Smoke		5 (5)	2 (4)	.76
Take vitamins		33 (34)	14 (29)	.58
Past research participant		48 (50)	10 (20)	.001

[a]Data are presented as total count and percent (intervention: N=99, control: N=49) unless otherwise noted. School year N=97 intervention, N=48 control. Transportation to campus N=98 intervention. Take vitamins: N=48 for control group. Past research participant: N=96 for intervention group.

[b]Student's t-test for age; all others are chi-square analysis of differences between groups.

[c]SE: standard error of the mean

Anthropometrics

At baseline, 22% (33/148) were overweight/obese (BMI ≥25 mg/kg^2). No significant differences were detected between treatment groups for students meeting health guidelines at baseline (Table 2). Baseline anthropometric data did not differ significantly between groups (Table 3). With the exception of WHR, most met recommendations for body measures. Further analysis of the WHR data revealed that all of those with elevated WHR were female, and of the 83 females with elevated WHR, 25 had WC measures that were below the recommended cut-off for disease risk (≤88 cm). Mean body fat mass percent (FM%) by sex was 29 in females and 20 in males.

Diet and Fitness

At baseline, 88% of participants (130/148) reported consuming <5 servings of fruits and vegetables daily, 59% (87/148) consumed >10% of their kcal from saturated fat; however, 91% (135/148) met/exceeded 150 min/week of moderate-vigorous exercise. Prevalence of measured high blood pressure (18%, 26/148) was higher than self-reported blood pressure (0.7%, 1/148). No significant differences were detected between treatment groups for students meeting diet and physical fitness guidelines at baseline (Table 2).

Table 2. Percentage of college students meeting health recommendations at baseline.

Recommendation		Intervention n (%)[a]	Control n (%)[a]	*P* [b]
Anthropometrics				
	FM% [c] (female: <32, male: <22)	54 (55)	33 (66)	.30
	BMI[d] (18.8-25g/m^2)	75 (76)	40 (80)	.60
	Neck circumference (female: <34 cm, male: <37 cm)	75 (76)	37 (74)	.68
	Waist circumference (female: ≤88 cm, male: ≤102 cm)	79 (81)	46 (92)	.08
	Waist-to-Hip ratio (Female: <0.8, male: <1.0)	43 (44)	21 (43)	.91
Dietary intake				
	Saturated fat (<10% daily kcal)	38 (38)	23 (46)	.58
	Total fat (<30% daily kcal)	97 (98)	49 (98)	.99
	Sugar (<10% daily kcal)	56 (57)	25 (50)	.45
	Fruit/vegetable (>5 servings/day)	13 (13)	5 (10)	.93
Fitness				
	Blood pressure (<120/80 mmHg)	83 (84)	39 (78)	.35
	Exercise[e] (≥150 min/week)	88 (89)	47 (94)	.31
	VO_{2max} [f] (mL/kg/min)	56 (58)	34 (69)	.17

[a]Data are presented as total count and percent (intervention: N=99, control: N=49) except FM% (Intervention: N=96) and VO2max (intervention: N=94). Dietary intake data and exercise are self-reported.

[b]Chi-square analysis of differences between groups (*P*<.05).

[c]FM%: body fat mass percent

[d]BMI: body mass index

[e]Exercise: min/week of moderate to vigorous physical activity

[f]VO_{2max}: maximum oxygen utilization. Met: >35, >33, >45, and >42 mL/kg/min for 19-year-old female, 20-year-old female, 19-year-old male, and 20-year-old male, respectively [40].

Repeated measures ANOVA with BMI and sex as covariates revealed a marginally significant linear time by treatment interaction for percent of energy from saturated fat, $F_{1,372}$=3.94, *P*=.048 (Figure 2). This was true for ITT analysis and completers. This indicates that the increase in saturated fat intake by the control group (0.3 [SE 0.30] % of kcal) was different from the decrease in saturated fat intake by the intervention group (0.7 [SE 0.42] % of kcal), *P*=.048. No significant effect of BMI or sex was detected for these variables. Further analysis of the change in saturated fat intake according to the Dietary Guidelines for Americans (DGA) recommendation (<10% of kcal) revealed a significant linear interaction of time and meeting/not meeting recommendations for the intervention group (*P*<.001). Thus, the decrease in saturated fat intake in the intervention group was largely due to those whose intake was higher than recommended (Figure 3). Additionally, the mean saturated fat intake for completers in the intervention group did not meet the DGA guidelines at baseline but did reach DGA levels by study completion. In contrast, the three completers in the control group who consumed saturated fat in excess of DGA guidelines at baseline never achieved DGA levels (Figure 4). No significant treatment by time interactions were detected for dietary intake of sugar, or fruit and vegetables, or for anthropometric or fitness variables (Table 3).

No significant difference in consumption of fruit or non-fruit snacks at baseline (*P*=.22) and week 12 visits (*P*=.06) was detected between the intervention and control groups. At week 24, significantly different consumption of fruit and non-fruit snacks ($\chi^2_{1,\ N=221}$ = 11.7, *P*<.001) was detected between the intervention and control groups (Figure 5).

Table 3. Body measures, dietary intake, and physical fitness of college students over time by treatment.

Variable		Baseline[a]		Week 12[a]		Week 24[a]		
		Int	Cont	Int	Cont	Int	Cont	*P*
Anthropometrics								
	FM%[b]	24.3 (0.50)	25.0 (0.68)	24.9 (0.50)	25.2 (0.68)	24.6 (0.50)	24.9 (0.68)	0.36
	BMI[c] (kg/m^2)	23.1 (0.38)	22.8 (0.54)	23.1 (0.38)	22.8 (0.54)	23.2 (0.38)	22.8 (0.54)	0.80
	NC[d] (cm)	34.4 (0.14)	34.4 (0.19)	34.4 (0.14)	34.4 (0.19)	34.5 (0.14)	34.6 (0.19)	0.34
	WC[e] (cm)	82.2 (0.49)	79.9 (0.68)	81.7 (0.49)	80.4 (0.68)	81.9 (0.54)	80.1 (0.75)	0.41
	WHR[f]	0.85 (0.004)	0.84 (0.006)	0.85 (0.004)	0.84 (0.005)	0.85 (0.004)	0.84 (0.006)	0.21
Dietary intake								
	Saturated fat[g] (% total kcal)	8.2 (0.29)	7.2 (0.41)	8.0 (0.32)	7.5 (0.42)	7.8 (0.34)	8.1 (0.45)	0.14
	Sugar (% total kcal)	10.6 (0.84)	12.1 (1.2)	10.2 (0.84)	12.0 (1.1)	10.1 (0.84)	10.6 (1.2)	0.32
	Fruit/veg[h] (cups/day)	2.6 (0.17)	2.7 (0.23)	2.4 (0.14)	2.3 (0.20)	2.4 (0.14)	2.4 (0.20)	0.64
Fitness								
	SBP[i] (mmHg)	111 (0.72)	111 (1.2)	111 (0.89)	111 (1.2)	110 (0.89)	110 (1.2)	0.92
	DBP[j] (mm Hg)	71.9 (0.72)	70.8 (1.0)	71.8 (0.72)	70.7 (1.0)	71.0 (0.72)	70.2 (1.0)	0.80
	Exercise[k] (min/week)	795 (58)	844 (80)	736 (58)	705 (80)	788 (58)	801 (80)	0.63
	VO_{2max}[l] (mL/kg/min)	41.4 (0.59)	42.6 (0.81)	41.1 (0.44)	42.1 (0.59)	40.8 (0.47)	42.4 (0.63)	0.83

[a]Int = Intervention. Cont = Control. Overall N=99 Int, N=49 Cont except FM% N=96 for Int, and VO2max N=94 for Int. Mean (standard error) derived by ANOVA repeated measures intent-to-treat analysis, adjusted for sex and baseline BMI.

[b]FM%: Fat Mass%

[c]BMI: body mass index

[d]NC: neck circumference

[e]WC: waist circumference

[f]WHR: waist-to-hip ratio

[g]Linear time by treatment interaction: $F_{1,372}=3.94$, $P=.048$

[h]Fruit/veg: fruit and vegetable

[i]SBP: systolic blood pressure

[j]DBP: diastolic blood pressure

[k]Exercise: minutes per week of reported moderate-vigorous activity

[l]VO_{2max}: maximum oxygen utilization

Figure 2. Saturated fat intake of college students over time by treatment presented as mean and standard error bars. Linear time by treatment interaction was significant (P = .048).

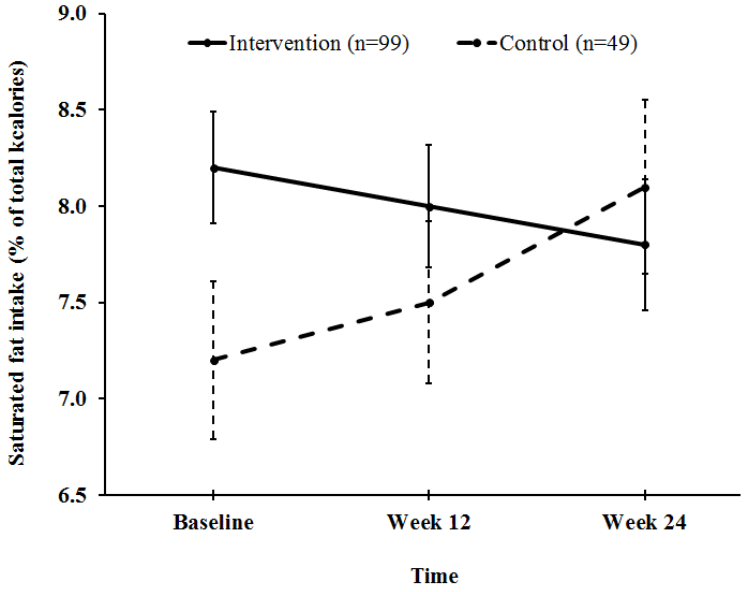


Figure 3. Change in saturated fat intake by college students in the intervention group who completed all study visits, categorized by met/not met recommendation at baseline, presented as mean and standard error bars. Slope of not met was significantly different than slope of met (P <.001).

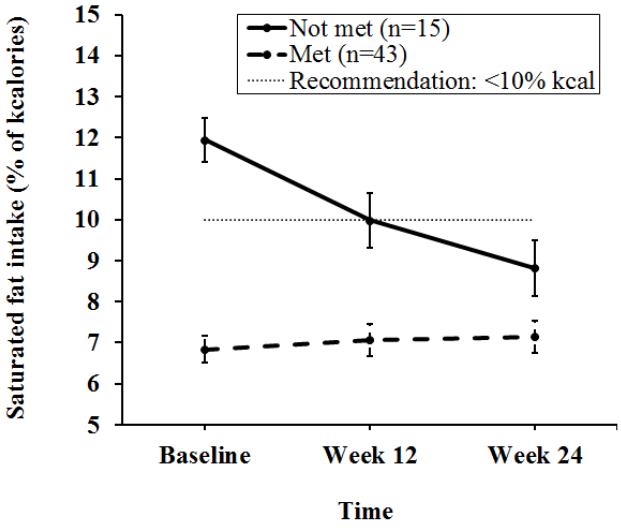


Figure 4. Change in saturated fat intake by college students in the control group who completed all study requirements, categorized by met/not met recommendation, presented as mean and standard error bars.

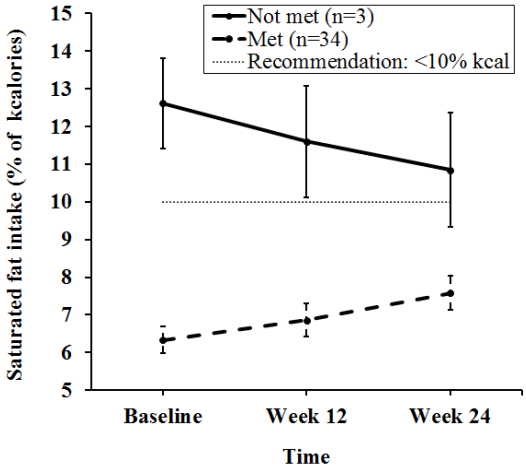
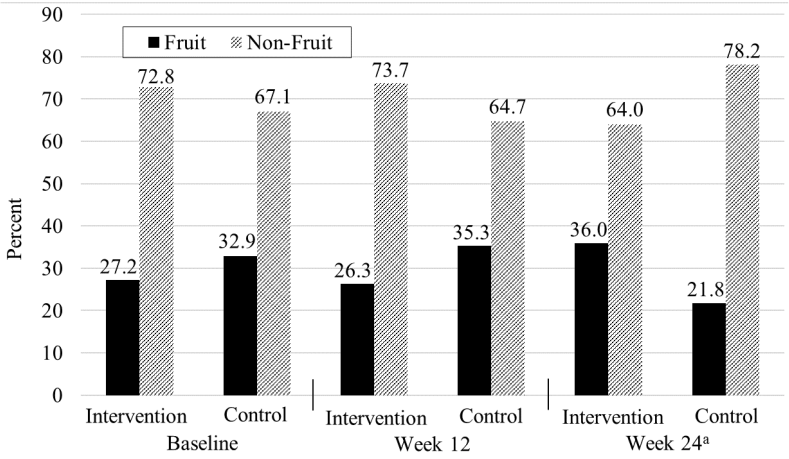


Figure 5. Percentage of fruit and non-fruit snacks consumed by college students. Significant difference was observed between treatment groups at week 24 only (P < .001).



Intervention

Among the 99 students in the intervention group, 32 chose goals to reduce fat and sugar intake, 32 chose to improve fruit and vegetable intake, and 35 chose to focus on physical activity. Of the 109 students who answered the satisfaction survey at week 24, 91 (84%) reported reading at least half of the study emails (control: n=35, intervention: n=56).

Discussion

Principal Findings

This study demonstrated the feasibility of ALIVE in a college population, by exceeding the recruitment goal with 106 participants who completed the study, a majority of whom reported reading most of the emails. ALIVE was effective in decreasing consumption of saturated fat and increasing the frequency of choosing fruit as a snack by a diverse sample of college students.

Intervention

Similar to other eHealth interventions in college students, small improvements in dietary intake but no changes in body measures were seen in the current study [26,28]. Differences between treatment groups in dietary intake of sugar, energy, vegetables, or for anthropometric or fitness variables were not detected. However, attenuation of BMI, WC, NC, WHR, body fat, blood pressure, sugar intake, and VO_{2max} in this study indicated that student health habits had not worsened. As seen in Table 2, large percentages of students required improvements in dietary sugar, fruit/vegetable, and saturated fat intake, whereas most students met requirements for body measures and physical activity. Additional studies are needed to assess the efficacy of eHealth in reinforcing optimal behaviors, particularly for those who do not meet health recommendations.

That the intervention group was not more successful compared to the control in attenuating health habits may arise from the insufficient power to detect these treatment effects, and the bias of repeated diet and physical activity surveys and body measurements in both treatment groups. According to previous studies, college students who are measured but receive no feedback, education, or intervention, show an increase in BMI, FM%, WC, and dietary fat intake, and decreased vegetable and fruit intake over time [9,10,41]. In the present study, control participants also received feedback and were encouraged to select a goal at baseline. Additionally, students in the intervention group were able to switch between the fruit and vegetable, fat and sugar, and physical activity health behavior tracks at any time. Thus, it is likely that only some students had the benefit of the full program of coaching. This could have weakened the effect, requiring a much larger sample size. It should also be noted that week 12 visits coincided with fall semester finals, which can be a high-snack, low-activity period in college life.

Anthropometrics

Although some eHealth programs were able to prevent increases in weight gain by college students, we were not able to support this finding [25,27]. However, the current study did demonstrate the importance of screening for high blood pressure in college populations and the importance of carefully selecting anthropometric measurements in order to detect disease risk. This study is the first to report NC in a diverse college sample [41,42]. A relatively unused measure, NC has been associated with obesity and cardiovascular disease risk [31,43]. In the current sample, approximately 25% of students were categorized as obese using NC, which is comparable to that identified by BMI. WC and WHR are often used to assess central obesity and are predictive of insulin resistance [44] and cardiovascular disease [41,45,46]. In our sample, 30% of females with elevated WHR had WC measures in the healthy range, indicating that WHR may not be a reliable indicator of central adiposity in a college-aged sample. NC is less intrusive than WC and should be further explored as an indicator of adiposity and disease risk in college students.

The students who failed to complete the study had significantly higher indicators of overweight/obesity, although only NC was clinically significant (higher and over recommendation) compared to those who dropped out. This may indicate that the program may not be well received by those with higher body weight.

Diet and Fitness

Less than half of the participants met the dietary guidelines for saturated fat intake at baseline, whereas the amount of total fat consumed met recommended levels. Thus, college students could benefit from interventions that alter food choices to improve the fat profile. The ALIVE program used in this study was designed to encourage and measure dietary changes in fat, sugar, fruits, and vegetables. We detected a linear time by treatment interaction, in which saturated fat intake worsened for the control group compared to the intervention group. Considering that the dietary guideline is 10% or less of calories from saturated fat [47], the observed difference in intake between groups of 1% of calories is clinically important.

Only 6% of college students report eating at least five servings of fruits and vegetables daily [14]. This is further supported by the current study, in which less than 15% of participants met the dietary guideline for fruit and vegetables. Snacks are a convenient way to bolster fruit and vegetable intake. Many are "self-packaged" and easy to carry (eg, oranges, apples, bananas). Given the limits of self-reported data, consumption of fruit as a snack was objectively measured in this sample of college students. Improved selection and consumption of fruit when offered as a snack was found after 24 weeks of the ALIVE program, whereas self-reported data showed no change in fruit and vegetable intake.

Lack of fitness as indicated by VO_{2max} and poor dietary patterns may explain the large portion of students having excessive weight and body fat. For example, almost half of the students in our study exceeded the World Health Organization recommendation for sugar intake, while a majority of students did not meet DGA guidelines for consuming fruit and vegetables. Dietary intake in this sample is consistent with body measures that indicate excessive weight in about a quarter of students studied.

In the current study, physical fitness was assessed by the Queen's step test, which estimates VO_{2max}. Categories of physical fitness as determined by VO_{2max} have been established by sex and age [40]. Higher numbers for VO_{2max} indicate better physical fitness. Overall, mean VO_{2max} placed the student sample in the "good" category for physical fitness level at baseline. Sixty-two percent of students had optimal physical fitness, which differs from the survey findings in which 90% of students reported engaging in the recommended 150 min/week of moderate to vigorous physical activity. Overreporting may explain the difference between the survey results and the fitness test results.

Limitations

As with most health behavior studies, self-selection bias of the convenience sample was a limitation in this study. Students who are interested in health are motivated to participate and may be more likely to improve health habits than others. The additional research experience in the intervention group could also have contributed to reporting bias between treatment groups.

Another major limitation involved exposure to feedback and promotion of health goals in the control group after the initial diet and physical activity surveys. Future studies could include a waitlist control or manipulate the initial survey to exclude feedback and goal selection.

Anthropometric measurements may be subject to some amount of error. For example, menstrual cycles may affect body measurements. We did not account for these potential confounding variables during the study.

Reported intake is known to be biased, with underreporting directly associated with BMI [48,49]. ALIVE used food frequency questionnaires to identify intake. Food frequency questionnaires are more biased and less correlated than food diaries [49]. Actual measurement of food intake is costly and time consuming. This study included the novel method of exploring the use of objectively measured partial dietary intake by measuring snack intake at study visits.

The fact that participants could choose very different goals (ie, increasing fruit/vegetable intake, decreasing fat/sugar intake, or increasing physical activity) and could switch tracks during the program indicated that we did not have sufficient power. Goal switching as a limitation has been addressed in ALIVE-PD, a future version of ALIVE, which is currently being evaluated for efficacy in adults. In ALIVE-PD, users work on diet and physical activity goals simultaneously, rather than switching between goals. Larger studies using ALIVE-PD in college students are warranted to document changes in diet and physical activity.

Conclusions

ALIVE is feasible in a diverse college population, as demonstrated by high participation and retention. Furthermore, health behaviors of college students, such as reducing saturated fat intake and increasing fruit intake can be influenced by ALIVE. Large randomized control trials using adaptations of ALIVE are warranted.

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Authors' Contributions

AS, CK, EM, and KL conceived the study. AS and JR conducted the study visits. AS analyzed the data and was the primary author.

Conflicts of Interest

None declared.

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Abbreviations

ALIVE: A Lifestyle Intervention Via Email
BIA: bioelectrical impedance analysis
BMI: body mass index
CCAPQ: Cross-Cultural Activity Patterns Questionnaire
DBP: diastolic blood pressure
DGA: Dietary Guidelines for Americans
eHealth: electronic health interventions
FM%: body fat mass percent
IRB: Institutional Review Board
ITT: intent-to-treat
NC: neck circumference
REDCap: Research Electronic Data Capture
SBP: systolic blood pressure
VO2max: maximum oxygen utilization
WC: waist circumference
WHR: waist-to-hip ratio

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Original Paper

Collecting Biospecimens From an Internet-Based Prospective Cohort Study of Inflammatory Bowel Disease (CCFA Partners): A Feasibility Study

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Abstract

Background: The Internet has successfully been used for patient-oriented survey research. Internet-based translational research may also be possible.

Objective: Our aim was to study the feasibility of collecting biospecimens from CCFA Partners, an Internet-based inflammatory bowel disease (IBD) cohort.

Methods: From August 20, 2013, to January 4, 2014, we randomly sampled 412 participants, plus 179 from a prior validation study, and invited them to contribute a biospecimen. Participants were randomized to type (blood, saliva), incentive (none, US $20, or US $50), and collection method for blood. The first 82 contributors were also invited to contribute stool. We used descriptive statistics and t tests for comparisons.

Results: Of the 591 participants, 239 (40.4%) indicated interest and 171 (28.9%) contributed a biospecimen. Validation study participants were more likely to contribute than randomly selected participants (44% versus 23%, $P<.001$). The return rate for saliva was higher than blood collected by mobile phlebotomist and at doctors' offices (38%, 31%, and 17% respectively, $P<.001$). For saliva, incentives were associated with higher return rates (43-44% versus 26%, $P=.04$); 61% contributed stool. Fourteen IBD-associated single nucleotide polymorphisms were genotyped, and risk allele frequencies were comparable to other large IBD populations. Bacterial DNA was successfully extracted from stool samples and was of sufficient quality to permit quantitative polymerase chain reaction for total bacteria.

Conclusions: Participants are willing to contribute and it is feasible to collect biospecimens from an Internet-based IBD cohort. Home saliva kits yielded the highest return rate, though mobile phlebotomy was also effective. All samples were sufficient for

genetic testing. These data support the feasibility of developing a centralized collection of biospecimens from this cohort to facilitate IBD translational studies.

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KEYWORDS

inflammatory bowel disease; biobank; Internet cohort; CCFA Partners

Introduction

Inflammatory bowel disease (IBD), including Crohn's disease (CD) and ulcerative colitis (UC), affects 1.1-1.4 million individuals in the United States and is increasing in prevalence [1,2]. IBD imparts significant morbidity to patients [3] and burden to the health system [4,5]. The pathogenesis of IBD is related to a combination of genetic susceptibility, environmental factors, and host-microbial interactions in the gut [6,7]. Recent genome-wide association studies reveal at least 163 susceptibility loci for IBD [8], emphasizing the range and complexity of pathways that may be involved.

Despite this emerging knowledge, little is known about how these factors impact disease risk [9] and even less about disease course and exacerbations. Such knowledge is necessary to define prognosis and response to treatment, guide medical decision making and lifestyle modifications, and ultimately lead to personalized medicine for IBD. In fact, the recent Crohn's and Colitis Foundation of America (CCFA) position paper on challenges in IBD identified studies to address these concepts as a top research priority [10].

Although case-control studies have historically been used for gene-environment studies, prospective cohort studies have many advantages, including the ability to study multiple outcomes [11] and critical evaluation of biological predictors of those outcomes. In fact, large prospective cohort studies with centralized biospecimen collection processes are considered "indispensable" by leaders in the field [12]. The Internet has the potential to be used to conduct gene-environment research remotely and at low cost with enhanced flexibility and rapidity, but to date it has not been widely utilized for these types of studies [13]. With the recent growing success of Internet-based cohorts and survey research [14-17], an opportunity to expand these cohorts to include biospecimen collection for gene-environment studies has now emerged.

CCFA Partners is an Internet-based cohort of over 13,000 adults with IBD that was developed in 2011 to accelerate clinical and patient-reported outcomes research [14]. Since its establishment, this cohort has been used in a number of cross-sectional and longitudinal studies covering a wide range of topics [10,14,18-22]. CCFA Partners has the potential to facilitate gene-environment and other translational studies, as well, if the cohort members would be willing to contribute biospecimens for molecular, genetic, and microbiological research. We previously surveyed over 1000 cohort members about their attitudes regarding biobanking, and an overwhelming majority (>90%) indicated willingness to contribute biospecimens [23]. However, little research exists on the practical aspects of collecting genetic or biospecimen samples from patients involved with Internet cohort studies.

Here, we report the feasibility of collecting saliva, blood, and stool from members of the CCFA Partners cohort in a systematic fashion for use in future studies. If feasible, this collection could provide a tremendous resource for IBD research and serve as a model for future methods of Internet-based translational research.

Methods

CCFA Partners

Methods for recruitment and prospective follow-up of participants in CCFA Partners have been previously described [14]. Inclusion criteria are ≥18 years of age, self-reported IBD, and Internet access. Participants complete a baseline survey upon registration and follow-up surveys every 6 months.

Biospecimen Collection

Our study was designed to collect and analyze approximately 100 blood samples (50 by mobile phlebotomist and 50 drawn through physician offices) and 100 saliva samples. A total of 179 CCFA Partners participants who previously participated in a validation study [18] ("Validated population"), in which their physicians were contacted to confirm their IBD type and characteristics, were randomized to each of the three specimen categories (blood by mobile phlebotomist, blood at physician's office, or saliva). Participants were also randomized to incentive level (none, US $20, or $50).

In addition to the validation cohort, we also randomized all CCFA Partners participants ("General CCFA Partners population") taking any survey between August 20, 2013, and January 4, 2014, according to the same study arms. Within each arm, participants were successively invited until the recruitment target was approached.

Consent forms described the purpose, potential impact, and potential risks of genetic studies on biospecimens, as well as privacy protections including de-identification of samples, physical lock-and-key of stored specimens, and encryption of all data. Consenting participants were mailed a biospecimen collection kit either to be sent back to the Biospecimen Processing Facility or contacted by the mobile phlebotomy service to schedule a time and location for blood draw, as applicable.

Our study was designed to collect 50 stool samples among participants who provided genetic specimens. To achieve this, the first 82 participants who submitted a blood or saliva specimen were then invited to contribute a one-time stool sample. Participants were compensated US $20 for stool

samples, regardless of whether they had been randomly assigned an incentive for the initial biospecimen.

For the mobile phlebotomy arm, we used Examination Management Services, Inc. (EMSI), a nationwide mobile specimen collection service. EMSI contacted participants to schedule a blood draw at a convenient time, and phlebotomists mailed blood samples directly to the Biospecimen Processing Facility per EMSI protocol. For the physician blood draw arm, we mailed each participant a kit containing blood draw supplies and a prepaid FedEx return label for overnight delivery. For the saliva collection arm, we mailed participants Oragene-500 oral collection kits (DNA Genotek, Inc.) with a prepaid FedEx Express saver return label. For stool, participants were instructed to ship stool samples on the day of collection with at least four -1 °C ice packs. All collection materials were affixed with a unique sample identification number and barcode, which was scanned when the specimen was processed by our lab.

Host Genetic Analysis

DNA was extracted from saliva samples using the Chemagic Magnetic Separation Module I (MSMI) robotic system (Perkin Elmer), using the Chemagic DNA Saliva Kit and the MSMI 24-rod head. The MSMI system isolated DNA after cell lysis via highly specific binding of the DNA to proprietary M-PVA magnetic beads. Once bound, the DNA was washed several times and then released from the magnetic beads. Optical density readings were taken on a Nanodrop to assess the 260/280 and 260/230 ratio quality metrics. DNA quantitation was assessed via Picogreen using the Quant-iT PicoGreen dsDNA Assay Kit cat# P7589 (Life Technologies). DNA was extracted from blood using Puregene high salt extraction chemistry on the AutopureLS DNA extraction robotic system. DNA quantitation and 260/280 and 260/230 ratio quality metrics were performed on a Nanodrop spectrophotometer.

Saliva and blood samples were genotyped for 14 IBD-associated single nucleotide polymorphisms (SNPs) using TaqMan SNP Genotyping Assays from Life Technologies. We used pre-designed assays for all but one SNP (rs2066847), for which a custom primer was designed using previously established sequences (Forward primer: GTCCAATAACTGCATCACCTACCT; Reverse primer: CAGACTTCCAGGATGGTGTCATTC Probe 1 - VIC-MGB; Dye: CAGGCCCCTTGAAAG Probe 2 - FAM-MGB; Dye: CAGGCCCTTGAAAG) [24]. Polymerase chain reaction (PCR) volume was 5 uL.

Fecal Microbial Analysis

Samples were aliquotted into cryovials and stored at -80 °C until the time of extraction. Bacterial DNA was extracted from 30-60 mg (solid) or 100-150 mg (liquid) of frozen fecal material as previously described [25]. Quantitative PCR was performed using primers for the 16S ribosomal ribonucleic acid (rRNA) gene of specific bacterial groups: forward, 5'-GTGSTGCAYGGYTGTCGTCA-3' and reverse, 5'-ACGTCRTCCMCACCTTCCTC-3', using 10 ng of DNA. Standard curves were generated using plasmids containing relevant PCR products for each bacterial group and used to enumerate copy number in individual samples.

Data Analysis

We used descriptive statistics and *t* tests or Fisher's exact test as applicable for comparisons between groups. All statistics were computed using SAS version 9.3. The study protocol was approved by the Institutional Review Board at the University of North Carolina at Chapel Hill.

Results

Study Population Characteristics

Of the 591 cohort member invited to contribute a biospecimen, 239 (40.4%) participants indicated interest and 171 (28.9%) contributed a biospecimen. In total, we collected 90 saliva samples, 47 blood samples from the mobile phlebotomy service, and 34 blood samples through physician offices. Demographic information for general CCFA Partners population included in this study and validated population participants is shown in Table 1. The general CCFA Partners population typically had lower education levels and a higher proportion of CD: 61.7% (254/412) versus 52.5% (94/179) CD for validated population. No significant differences were found across any other factors such as age, sex, race, or disease duration. Demographic factors for participants who indicated interest but did not contribute a specimen were compared to contributors (data not shown), and no significant differences were found.

Table 1. Study population characteristics stratified by random selection versus selection from prior validation study participants and by biospecimen contribution status.

	Selection status		General CCFA Partners population			Validated population		
	CCFA Partners general population (n=412)	Validated population (n=179)	Contributed (n=93)	Did not contribute[a] (n=319)	*P*	Contributed (n=78)	Did not contribute[a] (n=101)	*P*
Female, %	71.4	73.2	73	70.8	.67	74	72.2	.74
Age in years, mean	45.1	46.6	46.9	44.6	.49	48.2	45.4	.60
Race, n (%)					.54			.62
White	365 (94.8)	156 (94.5)	81 (94)	285 (95.0)		68 (96)	88 (93.6)	
Black/African American	8 (2.0)	4 (2.4)	1 (1)	7 (2.3)		1 (1)	3 (3.2)	
Asian	1 (<1.0)	1 (<1.0)	0 (0)	1 (<1.0)		0 (0)	1 (1.0)	
Other	11 (2.9)	4 (2.4)	4 (5)	7 (2.3)		2 (3)	2 (2.1)	
Education, n (%)					.82			.04
12th grade or less	24 (6.0)	4 (2.4)	4 (4)	20 (6.5)		0 (0)	4 (4.2)	
Some college	101 (25.6)	30 (18.0)	21 (24)	80 (26.1)		11 (15)	19 (19.8)	
College	162 (41.0)	73 (43.7)	39 (44)	123 (40.1)		28 (39)	45 (46.9)	
Graduate school	108 (28.0)	60 (35.9)	25 (28)	84 (27.4)		32 (45)	28 (29.2)	
Disease type, n (%)					.73			.14
CD	254 (61.7)	94 (52.5)	59 (63)	196 (61.4)		46 (59)	48 (47.5)	
UC/IC	157 (38.1)	84 (46.9)	34 (37)	123 (38.6)		32 (41)	52 (51.5)	
Disease duration in years, median	11.4	11.3	13	11.1		13	10.0	

[a]Includes participants who did not indicate interest and participants who indicated interest but never submitted a biospecimen.

Demographic Factors Associated With Biospecimen Return Rates

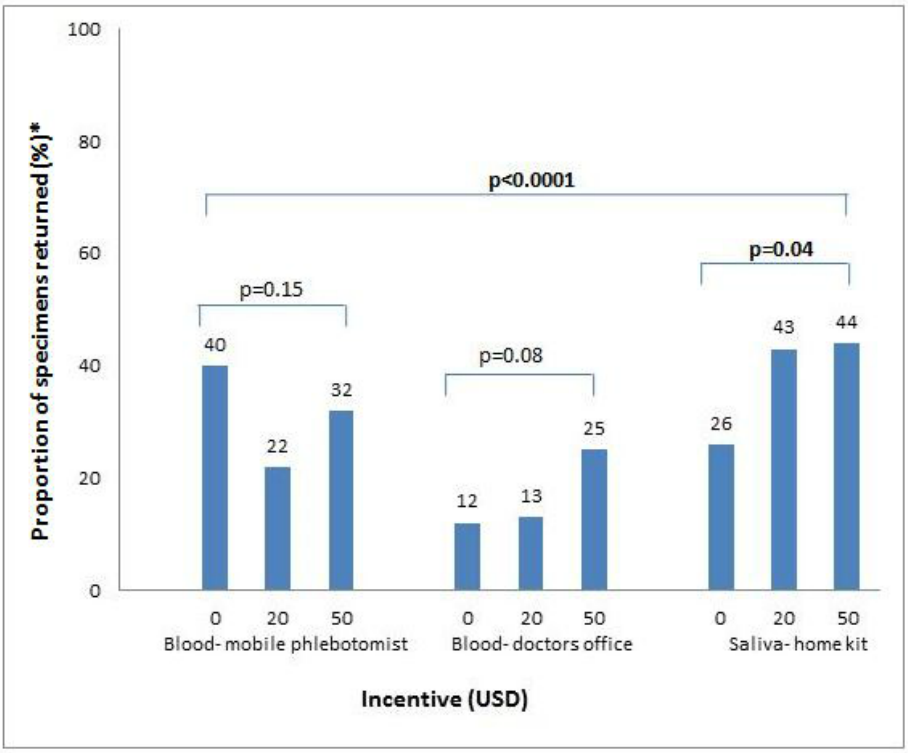
Overall, age, sex, race, disease type, or duration were not related to contribution status. Participants from the validated population were twice as likely to submit a biospecimen than general CCFA Partners population: 43.6% versus 22.6% (78/179 versus 93/412, respectively), *P*<.001. Within this subgroup, higher education level was significantly associated with contribution status (*P*=.04) as shown in Table 1.

Return Rates by Biospecimen Type and Incentives

A total of 171 participants contributed blood or saliva. Four additional participants attempted to contribute, but for process reasons these were not obtained or biospecimen type was switched, so they were excluded from return rate analysis. Among biospecimen types, the return rate for saliva was higher than blood collected by mobile phlebotomist and at the doctor's office (38%, 31%, and 17% respectively, *P*<.001) as shown in Figure 1. For saliva, US $20 and $50 incentive were associated with significantly higher return rate than no incentive: 43% (34/80) versus 26% (21/80), *P*=.03, and 43% (35/80) versus 26% (21/80), *P*=.05. For blood drawn at a doctor's office visit, incentives typically showed a higher return rate, particularly the $50 incentive, but this did not reach statistical significance (*P*=.08). For blood collected by mobile phlebotomist, monetary incentive was not associated with an increased return rate. Of participants who submitted blood or saliva, 60% (49/82) also submitted a stool sample. There were no significant differences in stool contribution rates across general CCFA Partners versus validated population status (data not shown).

Return rates for each method and level of incentive were stratified by sex, prior participation in validation study, and race and education level as a proxy for socioeconomic status. An effect of incentives for saliva was observed in males, with 23% return rate for no incentive (5/22), 47% (9/19) for $20, and 58% (15/26) for $50 (*P*=.045). For females, the highest return rate for saliva of 43% was achieved with $20 incentive (26/61) but this was not statistically significant (*P*=.22). For saliva collection in participants who identified as white race, the $20 incentive yielded the highest return rate of 47% (34/76, *P*=.01,). There were no other significant differences in return rate across sex, prior validation study participation status, race, or education level (data not shown).

Figure 1. Proportions of biospecimens returned by collection method and level of incentive.



*Defined as total number of biospecimens returned to biospecimen processing facility by number of email invitations sent

Host Biospecimen Genotyping

A total of 171 samples were received (90 saliva, 81 blood). For saliva, total DNA yield ranged from 2.13-158.12 ug (median 52 ug) and 87% (81/93) of the samples yielded >20 ug. For blood, total DNA yield ranged from 6.59-382.14 ug (median 159 ug), 94% (76/81) of the samples yielded >50 ug, and 83% (67/81) yielded >100 ug. All samples were genotyped for 14 single nucleotide polymorphisms (SNPs) associated with IBD and risk allele frequencies (RAFs) were calculated. For all SNPs, the RAFs observed in our population were comparable to those in other large IBD populations [8,26] as shown in Table 2. Individual SNP frequencies for IBD overall, and for CD and UC, are provided in Multimedia Appendix 1. Crohn's disease-associated SNPs like NOD2 (rs2066844, rs2066845, rs2066847) were more common in CD patients than UC patients. Of 2394 possible genotypes, 32 (1.3%) were undetermined. Of these undetermined genotypes, 53% (17/32) came from saliva and 47% (15/32) from blood samples.

Table 2. Risk allele frequencies for SNPs in the CCFA Partners cohort compared to other large IBD populations.

SNP	Notable genes	RAF	Reference[a]
rs12994997	ATG16L1	0.58	0.52
rs6426833		0.56	0.54
rs6017342	ADA,HNF4A	0.52	0.53
rs11209026	IL23R,IL12RB2	0.98	0.93
rs3024505	IL10,IL20,IL19,IL24; PIGR,MAPKAPK2; FAIM3,RASSF5	0.15	0.16
rs10761659		0.62	0.54
rs2155219		0.52	0.51
rs1893217		0.18	0.16
rs2413583	ATF4,TAB1, APOBEC3G	0.89	0.83
rs11564258	LRRK2,MUC19	0.03	0.03
rs2066844	NOD2	0.05	0.07[b]
rs2066845	NOD2	0.05	0.02[b]
rs2066847	NOD2	0.05	0.02

[a]RAF values obtained from [8].

[b]RAF values obtained from [26].

Fecal Microbial Analysis

A total of 49 stool samples were received. Of these, 18% (9/49) were liquid stool. Total bacterial content ranged from 6.04x10^2 to 4.97 × 10^6 16S sequences/mg stool, as shown in Table 3. Characteristics of individual stool samples are shown in Multimedia Appendix 2.

Table 3. Bacterial content of stool samples.

	Total bacteria, 16S sequences/mg stool (n=49)
Minimum	604
25% percentile	111,400
Median	436,000
75% percentile	683,500
Maximum	4,970,000
Mean	557,554
Standard deviation	754,369
Standard error of mean	107,767
Lower 95% CI of mean	340,874
Upper 95% CI of mean	774,234

Discussion

Principal Findings

These data show that participants from an Internet-based IBD cohort are willing to contribute, and it is feasible to collect, biospecimens in a centralized fashion for use in translational research. The highest return rates were obtained from home saliva kits, though a mobile phlebotomy service was also effective for collecting blood samples. Among study participants who contributed blood or saliva, stool collection is also feasible. All biospecimens collected provided sufficient quantity and quality of material for genetic or microbiological analysis. As over 6000 CCFA Partners participants complete 1 or more surveys each year, we estimate that, if taken to scale, the cohort could collect >1800 biospecimens with a 1-year period. Taken together, these findings suggest that the CCFA Partners cohort is a valuable resource for future translational research studies.

CCFA Partners participants who previously participated in a study to validate IBD diagnosis [18] were significantly more likely to contribute a biospecimen than participants from the general CCFA Partners population. This is likely due to the fact that by participating in the prior study, they had demonstrated

that they were highly engaged research participants. Higher levels of education were associated with higher return rates within this subset, as well, indicating that there may be a particularly educated and motivated subset of the CCFA Partners cohort.

Our previous survey-based study of biobanking attitudes found that 39% of the surveyed cohort would "definitely" donate and 56% would "probably" donate biospecimens for research [23]. Our return rate of 29% out of all participants contacted for potential interest in this study is somewhat low in comparison. This discrepancy brings into question the validity and utility of hypothetical willingness surveys; however, differences in the response to a hypothetical and actual scenario are not entirely unexpected and practical or logistical concerns may have limited sample collection rather than lack of willingness. Findings from the willingness surveys could represent the highest proportion of participants that would contribute a biospecimen and thus could be used as a goal for overall rates of contribution. Additionally, our previous survey found that pharmaceutical funding negatively impacted stated willingness to contribute biospecimens [23]. As this pilot study was supported by industry, which was indicated on the consent form, this could also have negatively impacted our collection rates.

Our return rates for saliva were significantly higher than for blood or stool. A number of reasons could contribute to this finding. First, there may be a lower perceived burden of collecting saliva than blood or stool because it is self-collected, can be done at home, can be collected immediately, is not painful, and manipulation of saliva may seem cleaner, more hygienic, or more comfortable than the other options. Indeed, in our previous survey of perceptions of biospecimen collection, sample type preference favored saliva over blood or stool (94% versus 90% and 77%, respectively). As not all patients undergo routine bloodwork, this may explain the lower rates of DNA collection in the doctor's office blood draw arm, as compared to the other arms.

The authors are unaware of any other publications on feasibility of collecting biospecimens from entirely Internet-based prospective cohort studies such as CCFA Partners; however, there is one cross-sectional Internet-based study of the feasibility of collecting both survey-based and biospecimen data in an elderly Welsh population [13]. The response rate for those with Internet access was approximately 40%, which is equal to the percentage of our population that indicated interest in the study. The return rate for biospecimens in the Welsh study was 75% for buccal swab and 70% for dry blood, which is equivalent to our biospecimen return rate of 72% for those who indicated interest in the study. Regarding collection rates by method of sample collection, our findings are also consistent with a prospective Nurse's Health cohort study based in Denmark (not Internet-based) that reported a higher return rate for self-collected DNA samples (72-80%), either saliva or buccal cell samples, versus blood samples collected during an office visit (31%) [27]. Internet-based interventional studies have also met success with remote collection of biospecimens, reporting return rates of about 80% [28,29].

Our previous study on willingness to contribute biospecimens did not find that incentives were a reported motivator for participants [23]. In contrast, we found a significant effect of monetary incentive on saliva collection. We also found an effect of monetary incentive at the highest price point for blood collection with a doctor's office kit, but this did not reach statistical significance. In contrast, the highest return rate for blood collected by mobile phlebotomy was with no incentive. Our finding that incentives were significantly associated with increased return rates of self-collected saliva specimens but not blood specimens collected by mobile phlebotomist or at a doctor's office visit may represent a stronger effect of incentives on specimens that can be directly collected by participants. The discordant effects of monetary incentives on overall blood collection could suggest that participants who do contribute are intrinsically motivated, or that our degree of incentive was not high enough to overcome direct costs or perceived burden to the participants who did not contribute. Our findings are consistent with the results of a smoking cessation study with geographically dispersed participants in which the highest monetary incentive was associated with a higher return rate of self-collected buccal cell DNA biospecimens [30]. In a breast cancer genetics study, a small monetary incentive increased blood spot biospecimen return rates in breast cancer cases, but not controls [31], suggesting other factors that affect participation. Indeed, factors such as race [32,33], perceived trust [33], and chronic disease state [34] have been reported to affect participation in biospecimen research, although these findings are not replicated across different populations [35,36].

In all, monetary incentives at the highest price point may be a motivating factor for contributing biospecimens in the CCFA Partners cohort. Other patient-level factors such as demographics, chronic disease state, trust, and intrinsic motivation may play a more important role. For future studies, the cost-effectiveness of incentives should be weighed against perceived motivation within a specific population.

Across all modalities of biospecimen collection (home collection kits for saliva, mobile phlebotomy and doctor's office kits for blood), we were able to obtain sufficient quantity and quality of genetic material for genetic analysis. Additionally, the SNP genotyping results show that the CCFA Partners population is representative of a large number of loci of interest in IBD research. These findings replicate previously established risk allele frequencies and known SNP associations, further supporting the utility of the CCFA Partners cohort for future genetic and translational studies. Stool samples in both solid and liquid form were sufficient for quantification of bacterial DNA and likely would be useful for microbiological and environmental studies of IBD.

Strengths and Limitations

CCFA Partners has many strengths including the large size, prospective design, and entirely Internet-based platform, which allows for the largest known sample size for collecting patient-reported data in IBD. The prospective design also allows us to link patient-reported data, biospecimens, and biospecimen-derived data to future outcomes. Strengths specific to this biospecimen feasibility study include randomization

across multiple strata including biospecimen type and incentive level and inclusion of all participants regardless of age or geographic location. Although we did target cohort members who previously participated in a study to validate IBD diagnosis, and therefore are more likely to be engaged and participate in this study, we analyzed return rates separately to eliminate selection bias. This group has now provided us with a repository of genetic and microbiological material in addition to detailed physician-validated information about their disease diagnosis, phenotype, and surgical history, which could be used for a variety of future translational research studies.

One limitation of this study is the relatively small sample size; however, this project was intended as a pilot and feasibility study. Nevertheless, there remains a possibility that larger numbers and greater statistical power would unmask other patterns in return rates, including differences by age, sex, race, disease type or disease duration, and the effect of incentives. While only four contributed biospecimens could not be obtained due to process factors (representing 1% of the sample size), this could represent a significant number or cost if biospecimens were to be collected on a much larger scale. By design, we attempted stool collection only among patients who provided genetic samples. While this allowed us to most efficiently estimate the proportion of participants who would provide both genetic and stool samples (an increasingly important aspect of translational IBD research), it did not allow estimation of the proportion of participants that would provide stool samples alone. Last, although CCFA Partners is a large IBD cohort and diagnoses have been validated [18], members tend to be highly educated and motivated, so these findings may not be generalizable to different IBD or other chronic disease populations.

Conclusions

In conclusion, the successful collection and analysis of biospecimens from the CCFA Partners Internet-based cohort represents a tremendous opportunity for a wide scope of IBD research, including genetic, molecular, microbiological, epidemiological, clinical, and outcomes studies. Platforms such as CCFA Partners may provide important opportunities to translate basic science knowledge into clinically useful information, leading the way

toward precision medicine.

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Authors' Contributions

RLR was involved with study concept and design, analysis and interpretation of data, drafting, and critical revision of the manuscript. ASG was involved with study concept and design, data acquisition and analysis, and critical revision of the manuscript. SFC was involved with study concept and design and critical revision of the manuscript. CFM was involved with study concept and design, statistical analysis and interpretation of data, and critical revision of the manuscript. WC was involved with computer programming and data acquisition and analysis. ELJ provided help with data acquisition and participant support for CCFA Partners. AAS collected and analyzed data. PB, HD, JL, and MG were involved with data collection, analysis, and critical revision of the manuscript. RSS was involved in study concept, critical revision of the manuscript, and study supervision and is the principal investigator of CCFA Partners. MDK was involved in all aspects of the study, including study concept and design, analysis and interpretation of data, critical revision of the manuscript, and study supervision.

Conflicts of Interest

MDK is a consultant to GlaxoSmithKline.

Multimedia Appendix 1

Supplementary tables.

[PDF File (Adobe PDF File), 11KB - resprot_v5i1e3_app1.pdf]

Multimedia Appendix 2

Total bacterial content of stool samples in the CCFA Partners cohort.

[PDF File (Adobe PDF File), 4KB - resprot_v5i1e3_app2.pdf]

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Abbreviations

CCFA: Crohn's and Colitis Foundation of America
CD: Crohn's disease
DNA: deoxyribonucleic acid
EMSI: Examination Management Services
IBD: inflammatory bowel diseases
PCR: polymerase chain reaction
RAF: risk allele frequency
SNP: single nucleotide polymorphism
UC: ulcerative colitis

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Original Paper

A Web-Based Intervention to Encourage Walking (StepWise): Pilot Randomized Controlled Trial

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Abstract

Background: Despite Internet-based interventions that incorporate pedometers with appropriate goal-setting processes and other theoretically-based behavior change strategies being proposed as a means of increasing walking behavior, few have incorporated all of these key features or assessed maintenance of behavior change.

Objective: The objective of our study was to investigate the effect of a 12-week pedometer step goal walking program individually tailored to baseline step counts, combined with an interactive support website for step counts, health parameters and motivation over 12 and 24 weeks.

Methods: Low active participants (mean [SD] 46.2 [11.2] years) were randomly assigned to the Stepwise (SW) intervention group (n=49) or a comparison (CP) group (n=48). SW received a pedometer, step goal walking program and access to the SW website (containing interactive self-monitoring and goal feedback tools, motivational messages and action and coping planning strategies). CP received a pedometer and locally available physical activity information. Step counts, BMI, resting heart rate, blood pressure and glucose, cholesterol and triglyceride levels, psychological well-being, perceived health, self-efficacy and self-determined motivation were measured at baseline, 12 and 24 weeks.

Results: Linear mixed model analysis found that both groups' step counts increased from baseline to week 12 (β = 11,002, CI 5739-16,266, *P*<.001) and 24 (β = 6810, CI 1190-12,431; *P*=.02). Group step counts were significantly different at week 24 with SW taking 8939 (CI 274-17604, *P*=.04) more steps compared to CP. Compared to baseline, both groups had improved triglyceride levels (0.14 mmol/L, CI -0.25 to -0.02, *P*=.02) at week 12, decreased diastolic blood pressure (4.22 mmHg, CI -6.73 to -1.72) at weeks 12 and 24 (3.17 mmHg, CI -5.55 to -0.78), improved positive (β = .21, CI 0.03-0.38, *P*=.02) and negative affect (β = -.15, CI -0.28 to -0.03, *P*=.02) at week 12, and perceived health at week 12 (β = 6.37, CI 2.10-10.65, *P*=.004) and 24 (β = 8.52, CI 3.98-13.06, *P*<.001). Total cholesterol increased at week 12 (0.26 mmol/L, CI 0.099-0.423, *P*=.006) and week 24 (0.38 mmol/L, CI 0.20-0.56, *P*<.001). Repeated measures ANOVA found motivation for walking improved from baseline with higher task self-efficacy (*P*<.001, η^2 = .13) and autonomous motivation (*P*<.001, η^2=.14) at weeks 12 and 24 and decreased controlled motivation (*P*=.004, η^2=.08) at week 24.

Conclusions: Both groups had similar improvements in step counts and physical and psychological health after 12 weeks but only the SW group successfully maintained the increased step-counts 24 weeks post-intervention. This suggests the step-goal based walking program combined with Internet-based behavior change tools were important for sustained behavior change.

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KEYWORDS

physical activity, behavior change strategies, pedometer, self-monitoring, step goal walking program

Introduction

Regular moderate-intensity physical activity amounting to at least 150 minutes per week is one of the best lifestyle choices an individual can make to improve health, sustain good health and prevent ill health [1-2]. Despite the widespread benefits of engaging in an active lifestyle, participation rates remain low in the (nonclinical) general population [3-4]. Walking is regarded as the modality most likely to increase activity levels [5-6] as it is safe, inexpensive, does not require any special clothes or skills, is accessible to all socio-economic groups and can be easily incorporated into a daily routine. Walking interventions are effective in improving health based parameters [7-10]. Research suggests that walking interventions that are tailored to the individual, incorporate the motivational function of a pedometer [6], are informed by behavior change theory [11] and utilize the power of a Web-based delivery [12] can successfully increase walking behavior.

Incorporating a pedometer into a walking intervention with a nonclinical population of adults is associated with significant increases in physical activity of 2000-2500 steps per day [13-15] and this increase has been associated with clinically relevant reductions in weight and blood pressure [14]. The association with other health variables which are risk factors for cardiovascular disease, such as cholesterol, triglyceride and glucose levels, remain inconsistent [10]. Reducing cardiovascular disease risk remains an important health outcome. Although there is a positive relationship between physical activity and cardiovascular health, understanding the ability of specific pedometer interventions to demonstrate changes in a variety of health variables remains important so that health care professionals are aware of the health benefits that can be accrued when they are promoting these interventions [10]. The success of pedometer-based interventions, and interventions delivered using the Internet, comes when they are informed by behavior change theory [10,16]. As suggested by Ritterband et al [17] it is unlikely that one single theory or model can explain behavior change in an Internet intervention. Consequently, this study is underpinned by a number of key behavior change techniques (BCTs) drawn from Social Cognitive Theory [18] and Self-Regulation Theory [19] known to effect change in walking behavior [16]. Self-monitoring and feedback strategies are important to increase awareness of behavior, provide a tangible record of success, instill accountability [20-21] as well as increase confidence and reduce perceived barriers to walking [22-23]. The motivational feedback provided should be tailored to participant characteristics rather than being generic [24]. Action planning and coping planning help translate intentions into behavior [25] and are important for relapse prevention [17]. Lastly, a goal-setting component is key to a successful pedometer intervention [11]. Goal-setting strategies used in pedometer studies vary considerably and range from a generic fixed goal of 10,000 steps per day [26], a fixed increment between 1000 steps per day to 3000 steps per day over baseline [27-28], to a 10-20% increase based on the previous weeks steps [26,29]. For inactive individuals to feel motivated and confident of being successful, goals need to be individualized so they are realistic, achievable and easily adjusted when necessary [11]. Consequently, we favor the approach used by Fitzsimons et al [30]. Their 12-week walking program gradually increased weekly step goals so that by Week 7 participants are aiming to achieve 3000 steps above baseline values at least 5 days per week. This equates to 30 minutes of physical activity and meets the physical activity recommendations for health [31]. Community-based participants following the walking program showed significant increases in step counts from baseline to 12 weeks [32] and to 12 months [33]. Positive affect and perceived health also improved. This walking program has not been tested using a Web-based delivery.

Pedometer-based interventions of varied length have been conducted using Internet technology [27-29,34]. The majority provide step goals and ask individuals to log steps, but few have incorporated more than 1 or 2 key BCTs suggested as important for successful step count increases. A meta-analysis has shown that the more BCTs incorporated into an intervention, the larger the effect on behavior [16]. As well as motivating behavior change, these techniques can reduce participant attrition and sustain engagement with an intervention [35-36] which is important as website visits, and therefore exposure and engagement with the intervention, have been shown to decrease over time [12]. One exception is Richardson et al's [28] 6-week intervention where participants with type 2 diabetes received tailored motivational messages (highlighting the benefits of exercise and how to overcome barriers), educational tips, automatically calculated goals and feedback in relation to their performance towards the goals. Results showed step counts increased by ~ 1950 steps per day. Our study differs from Richardson et al in that we are targeting a nonclinical sample of adults, our motivational messages target the building of both task and barrier self-efficacy as well as autonomous motivation, we offer a different approach to goal-setting and the study is longer in duration. Furthermore, Richardson et al's research, similar to most Web-supported studies, did not measure maintenance of step changes following the intervention. Carr et al [34] investigated maintenance of step counts and found step counts had returned to baseline after 8 months. However, this was not a pedometer intervention; they simply used pedometers to measure physical activity behavior. The importance of investigating sustained behavior change from a Web-supported pedometer intervention has been recognized [13,37].

To overcome the limitations of previous research, we created the StepWise intervention which combines an individually-tailored step goal pedometer walking program with an interactive support website. The website allows the participants to enter their step counts, graphically see their goal achievement, obtain automated and individualized motivational messages and plan their walking activities. This pilot study consisted of a 12-week intervention with a 12-week follow-up to investigate whether the StepWise intervention would increase

step counts, improve health parameters and motivation for walking compared with a comparison group in a community sample of apparently healthy adults.

Methods

Study Design

This pilot randomized trial compared a StepWise intervention group (SW) to a comparison group (CP) over a 12-week intervention and a 24-week follow up. The intervention was fully Web-based but the study also involved face-to-face components, specifically to collect outcome data and for the intervention and comparison procedures to be explained to participants. The trial is reported in accordance with CONSORT-EHEALTH guidelines. The research was approved by the University Human Ethics Committee in accordance with all applicable regulations (July 9th, 2012, reference 12/159).

Participants

A targeted recruitment strategy was employed [38] to attract participants from the community who were not currently meeting physical activity recommendations (ie, participated in <150 minutes of moderate intensity physical activity per week), who were aged over 25 years (to exclude a student population) and were apparently healthy. Participants were recruited offline in September of 2012. Advertisements were placed in community newspapers, a recruitment email was sent through the internal email systems of the local University, Polytechnic, City Council, primary and high schools, and posters were placed in areas where low-active individuals would see them (eg, GP practices, supermarkets, local shops, community and church halls). To be eligible, individuals had to be able to walk, have no contraindications to participate in a moderate-intensity walking program, and have regular access to the Internet (it was presumed that participants who responded to the recruitment advert would be computer/Internet-literate). Interested participants contacted the research team via email or telephone and were given detailed information about what was involved in study participation. To screen for eligibility, participants were asked to explain what physical activity they currently participated in and whether or not they were taking any medications for health conditions. Those participants who reported taking medication for blood pressure or cholesterol were admitted to the study, but their data was not used in the analysis of those variables. Those who met the study criteria and were still interested in participating attended a baseline testing session where they completed the Physical Activity Readiness Questionnaire [39] to ensure they had no contraindications to participate in physical activity and provided informed consent after the nature and possible consequences of the study were explained.

Measures

All measurements took place in a room at the University in the morning. Participants arrived in a fasted state having done minimal physical activity that morning. The same measurements were taken at baseline, 12 weeks, and 24 weeks.

Primary Outcome Measure: Step Counts

Physical activity was assessed by step counts. Participants were given a Yamax PW-610 pedometer, individually calibrated consistent with manufacturers' guidelines and asked to wear the pedometer during all waking hours for the next 7 days and to remove it only when sleeping, bathing or during water-based activities. The screen of the pedometer was covered so participants could not see their step counts. At baseline they were encouraged to continue with the same amount of physical activity as they had been doing the previous week. One week later, the researcher removed the screen cover and step counts were recorded. The pedometer was returned to the participant to use in the study.

Secondary Outcome Measures

Health Variables

Participants were measured for height and weight in order to calculate Body Mass Index (weight in kg/height in m^2), seated resting heart rate (measured using a Polar PE3000 heart rate monitor) and blood pressure (using a manual sphygmomanometer). A trained phlebotomist then drew a 6 ml blood sample from the participant's arm by venipuncture and the sample was analyzed for glucose, total cholesterol, HDL cholesterol and tryglycerides. After collection the venous blood was centrifuged and the heparinized plasma was analyzed for Glucose, Total Cholesterol, HDL cholesterol and Triglycerides using a Cobas C111 analyzer (Roche Diagnostics). Staff taking these measurements and doing the analysis were blinded to group allocation. Participants completed the Positive and Negative Affect Schedule [40] to assess psychological well-being (measured on a 5-point Likert-type scale, ranging from "very slightly or not at all" to "extremely"). Positive affect reflects the extent to which the individual feels enthusiastic, active and alert, a state of pleasurable engagement. Negative affect reflects the extent to which the individual experiences subjective distress and unpleasurable engagement that subsumes a number of aversive mood states. Positive and negative affect are two distinct dimensions of affective state and are not bipolar opposites [40]. Participants also completed the Visual Analogue Scale of the Euroqol EQ-5D [41] to assess self-rated health status (measured on a 100 point scale, from "worst health you can imagine" to "best health you can imagine").

Motivation

Participants completed a number of motivation questionnaires. The Behavioral Regulation in Exercise Questionnaire-2 [42] is a scale from which a measure of autonomous motivation (motivated by value attached to the outcomes of being active and the enjoyment gained) and controlled motivation (motivated by need for reward or as a result of feeling pressured to be active) was created [43-44]. Items are measured on a 5-point Likert-type scale ranging from "not true for me" to "very true for me", The Barriers for Habitual Physical Activity Scale [45] assessed the barriers to participating in physical activity (measured on a 5-point Likert-type scale ranging from "strongly disagree" to "strongly agree"). Finally, measures of self-efficacy for walking [46] and for overcoming barriers to exercise [47]

were measured on a 10-point scale from “not at all confident” to “completely confident”.

Intervention Use

Participants were encouraged to log into the website at least once a week. Website usage statistics were downloaded at week 12 to assess engagement and adherence to the intervention. Additionally, participants completed a questionnaire asking (1) whether they had used the SW website (yes/no), (2) how often (more than once a week, once a week, once every 2 weeks, 3-4 times over the 12 weeks, 1-2 times over the 12 weeks), and (3) to write down what aspects of the website they had found most and least useful.

Randomization

One week after baseline testing, participants attended a second face-to-face session where they were randomized into either SW or CP groups. To ensure equal representation in the groups, randomization was stratified by gender (male or female) and age (<45years or >45years) creating 4 distinct stratification groups. Group assignment was placed inside sealed envelopes and the envelopes shuffled to produce an unpredictable sequence of group assignment. When each participant arrived, the next envelope in the pile representing that individual (male or female and <45 or >45years) was opened to reveal their group assignment. As far as we are aware, participants could not tell whether they were in the SW or CP group because all study information stated participants would be given access to a pedometer walking program and a supportive physical activity website.

StepWise Intervention Group

The SW intervention consisted of 2 components: (1) an individualized pedometer-based walking program with weekly step goals, and (2) a website (created by the University Web development team) where individuals entered their step counts, received goal feedback, their next weekly goal (from the walking program) and tailored motivational feedback, and created a physical activity plan. At the second face-to-face session, the intervention components were discussed with each participant and the participant left with an information sheet summarizing the discussion. The same researcher met with each participant and the information discussed was standardized across all participants to ensure accuracy and consistency in the delivery of the intervention.

Individualized Pedometer-Based Walking Program

The walking program was structured around each participant’s baseline step counts and designed so that physical activity increased gradually [30,32-33]. By the seventh week, participants would be walking an extra 3000 steps per day over their baseline and meeting the physical activity guidelines (see Textbox 1).

Textbox 1. Pedometer-based incremental walking program goals.

- Week 1: Walk an extra 1500 steps (from baseline value) on at least 3 days of the week
- Week 2: Walk an extra 1500 steps (from baseline value) on at least 3 days of the week
- Week 3: Walk an extra 1500 steps (from baseline value) on at least 5 days of the week
- Week 4: Walk an extra 1500 steps (from baseline value) on at least 5 days of the week
- Week 5: Walk an extra 3000 steps (from baseline value) on at least 3 days of the week
- Week 6: Walk an extra 3000 steps (from baseline value) on at least 3 days of the week
- Week 7: Walk an extra 3000 steps (from baseline value) on at least 5 days of the week
- Week 8: Walk an extra 3000 steps (from baseline value) on at least 5 days of the week
- Weeks 9-12: Maintain walking levels using the week 7 goal

StepWise Website

Each participant’s baseline step counts and reported barriers to physical activity (from Barriers for Habitual Physical Activity Scale) [45] were entered manually by the researcher and an automated algorithm generated the individual’s step count goal for each week (based on the walking program). Each participant created their own website log-in and password. Participants were sent an email at the end of each week prompting them to log-in to the website and enter their weekly step counts (read from the pedometer memory). The website algorithm calculated whether or not they had achieved their weekly step goal and generated a motivational message relating to whether they had been successful or unsuccessful in achieving their goal (see Figure 1 for message examples). A graph showed the step goal number and the actual steps achieved. Above the graph, the step goal number for the next week and a motivational tip to help them achieve it was displayed (see Figure 2). These motivational messages and tips (which changed each week) were created based on behavior change theory around building task and barrier self-efficacy and autonomous motivation. Finally, participants were prompted to use the activity diary feature to write an action plan for how they would achieve their step goals and a coping plan to overcome any barriers they might face in trying to achieve that plan.

Figure 1. Examples of success and failure messages shown once step counts were entered.

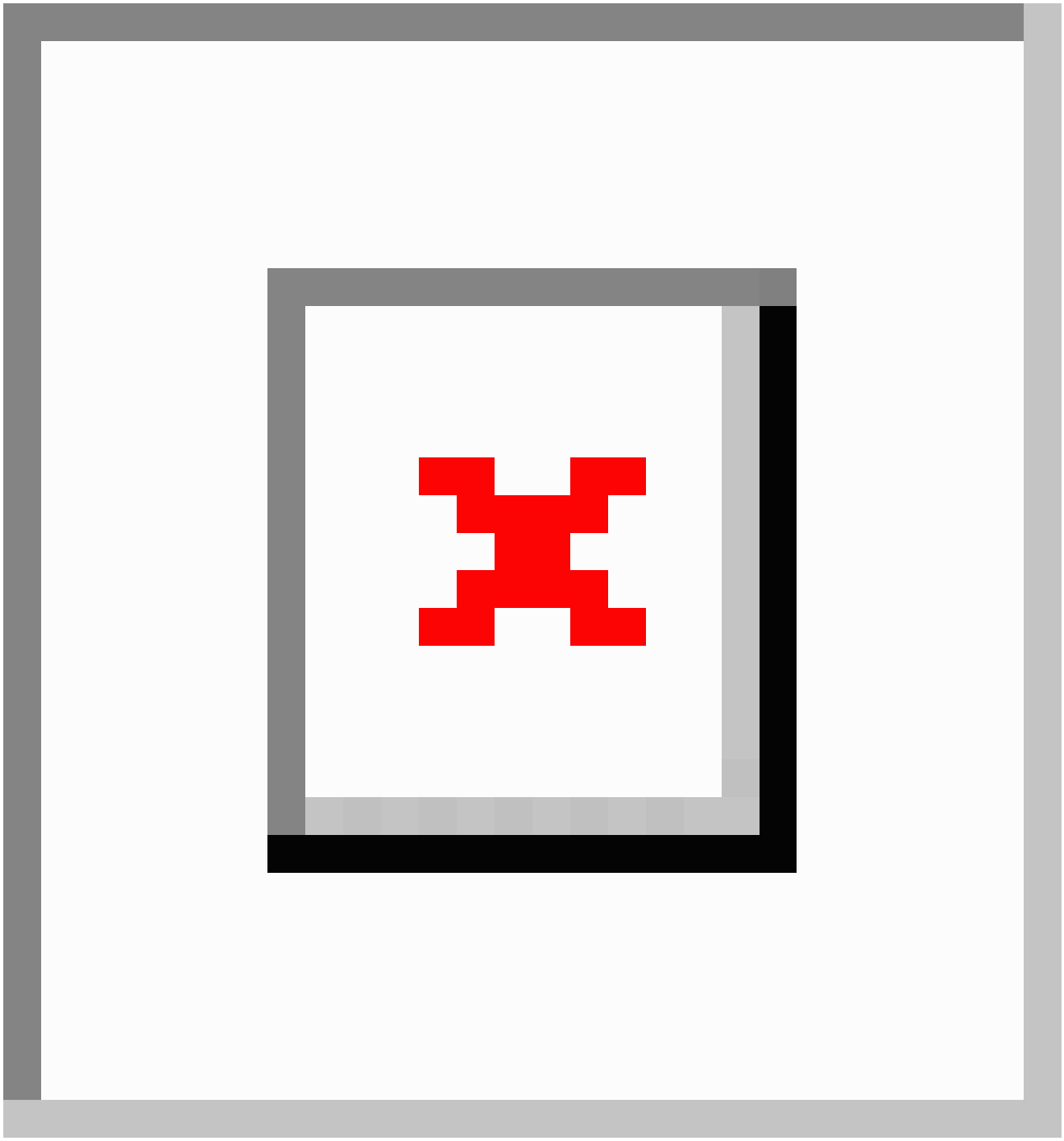
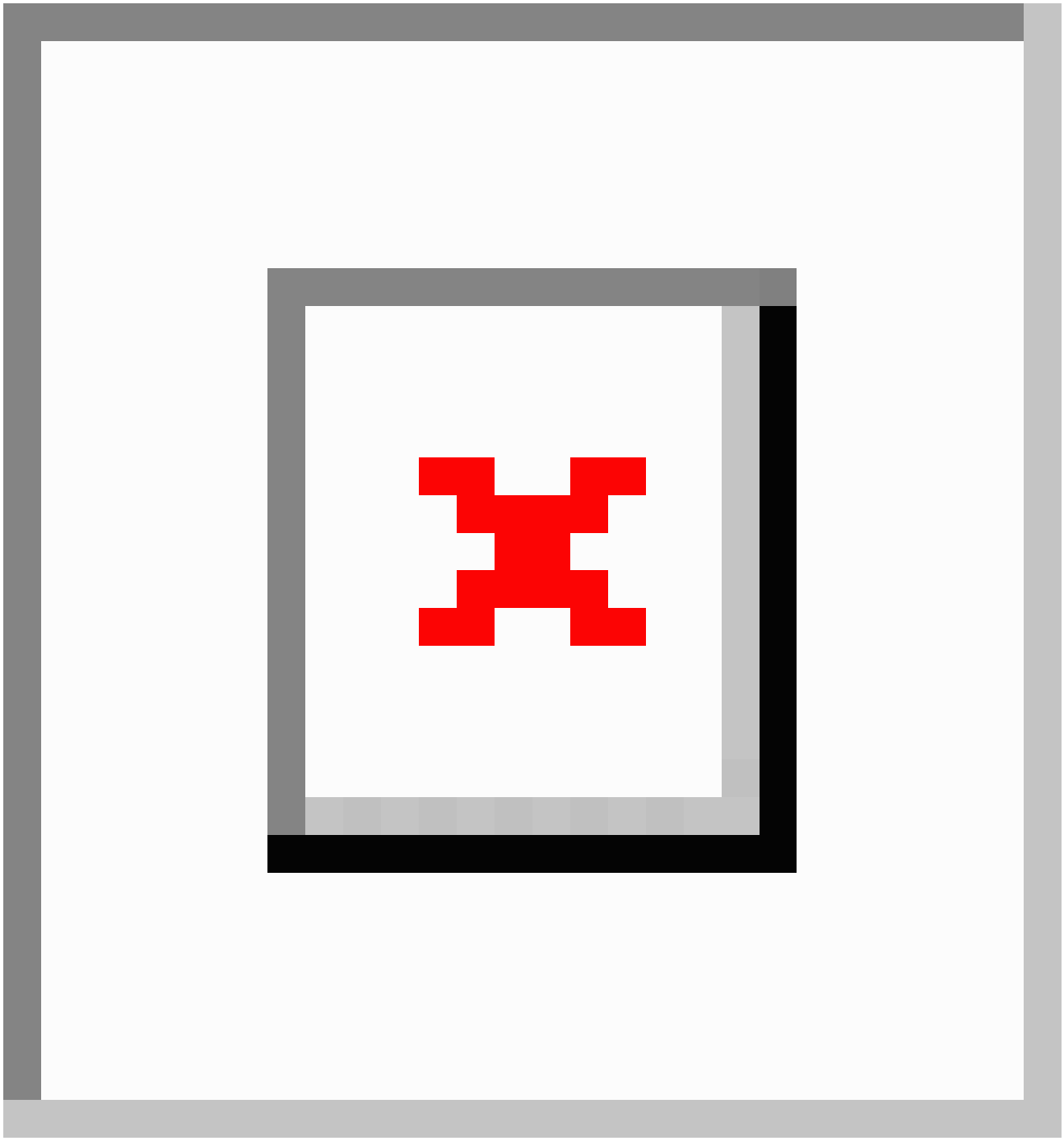


Figure 2. The graph showing step goals and number of steps achieved each week and the step goal for the following week along with a motivational message.



Twelve-Week Follow-Up

At the end of the 12-week intervention, participants met with the researcher and were encouraged to maintain their new level of physical activity over the following 12 weeks. They kept the pedometer and were told they could continue to access and use the StepWise website to receive step goals. To support maintenance of behavior change, there was a discussion of strategies relating to relapse prevention. This involved encouraging the participant to identify factors that could interfere with their being able to continue their new walking behavior and thinking of strategies they could put in place to overcome these factors so they could remain active. In the 12-week follow-up period, participants were sent 2 emails reinforcing the messages that had been discussed.

Comparison Group

Comparison group (CP) participants were given a pedometer and informed of the public health guidelines for physical activity (150 minutes of weekly moderate intensity physical activity) and how the guidelines translated into pedometer steps. Participants were shown the MoveMe website [48] a noninteractive city-specific physical activity website which provides information on physical activity and local physical activity opportunities. In comparison to the StepWise website, the MoveMe website does not contain any interactive features known to encourage physical activity behavior change.

Participants were encouraged to access the MoveMe website regularly and use the resources to help them become more active. Participants left with an information sheet summarizing the discussion. To ensure equal contact time with SW group, participants were sent a generic weekly email reinforcing the physical activity message and to access the MoveMe website for information. This comparison group was chosen over a minimal or no intervention control group because it reflects what could be considered standard practice and provides a test of how the SW intervention compares to this standard practice. The information and support given to the comparison group as well as their receiving a pedometer reflects what is currently available locally for individuals wanting to increase their physical activity.

12-Week Follow-Up

At the end of the 12 weeks, participants met with the researcher and were encouraged to maintain their new level of physical activity over the following 12 weeks, or to continue to try and meet the public health guidelines for physical activity and to continue to use the MoveMe website. Participants kept their pedometer to use during this period if they wished. In this follow-up period, participants were sent 2 emails reinforcing the messages that had been discussed.

Sample Size

G-Power analysis [49] was used to calculate sample size for between-group analyses of weekly step counts (the primary outcome measure) with repeated measures. Power was set at 0.8, alpha set at 0.05 with a medium effect size (Cohen's f=.25) expected based on the results of Baker et al [32] who utilized the same pedometer-based goal program. Assuming a correlation among repeated measures of 0.5 with 2 groups and 3 measurements per group, the required total sample size was 86 (43 per group).

Data Analysis

To ensure the groups were comparable, independent t-tests were used to analyze demographic variables and baseline data. The step counts and health variables were analyzed by linear mixed effect models using the R statistical package [50]. Group (2 levels: SW v CP) and time (3 levels: baseline, 12 and 24 weeks) were treated as fixed effects with time a repeated fixed effect. The baseline data and comparison group were used as the reference variable. Participant was a random effect. Several models were conducted (with interaction, without interaction, no random effect, etc) and Akaike Information Criterion comparison was used to assess the best model fit. The main effects and interactions were followed up using general linear hypothesis testing. The blood pressure, cholesterol and/or glucose data from participants who reported taking medications for high blood pressure, cholesterol or Type II diabetes were not used in the analysis of those variables. The motivation data were analyzed using time × group repeated measures ANOVA (MANOVA for barriers to being active) followed up by Bonferroni post hoc tests, the effect size for any effects is denoted by partial eta^2($\eta 2$).

Results

Participants

Of the 152 people who expressed interest and were eligible to participate, 103 attended baseline testing and 5 people were put on a waiting list (due to funding restrictions, we had a limit of ~100 participants). After baseline testing, 97 participants (82 women, 15 men) were eligible to be randomized, and 42 females and 7 males with a mean age of 47.1 years (SD 11.3) were allocated to the SW group while 40 females and 8 males with a mean age of 45.3 (SD 11.1) were allocated to the CP group (see Figure 3). Participant demographics are shown in Table 1. At baseline testing, 6 participants had to be excluded because they attended with a friend or family member and this compromised the ability to randomly allocate them to a group. Had they been allocated to different groups there would have been contamination between conditions due to the likelihood of them sharing information. Of those who took part in the study, 6 SW and 7 CP participants reported taking medication for hypotension, 1 CP and 1 SW participant were taking medication for high cholesterol and 1 CP participant reported having type 2 diabetes. There were no significant differences between the groups in any of the variables at baseline (see Table 1). Study requirements were completed by 33 SW and 34 CP participants and their data were included in the analysis.

Table 1. Participant baseline characteristics[a].

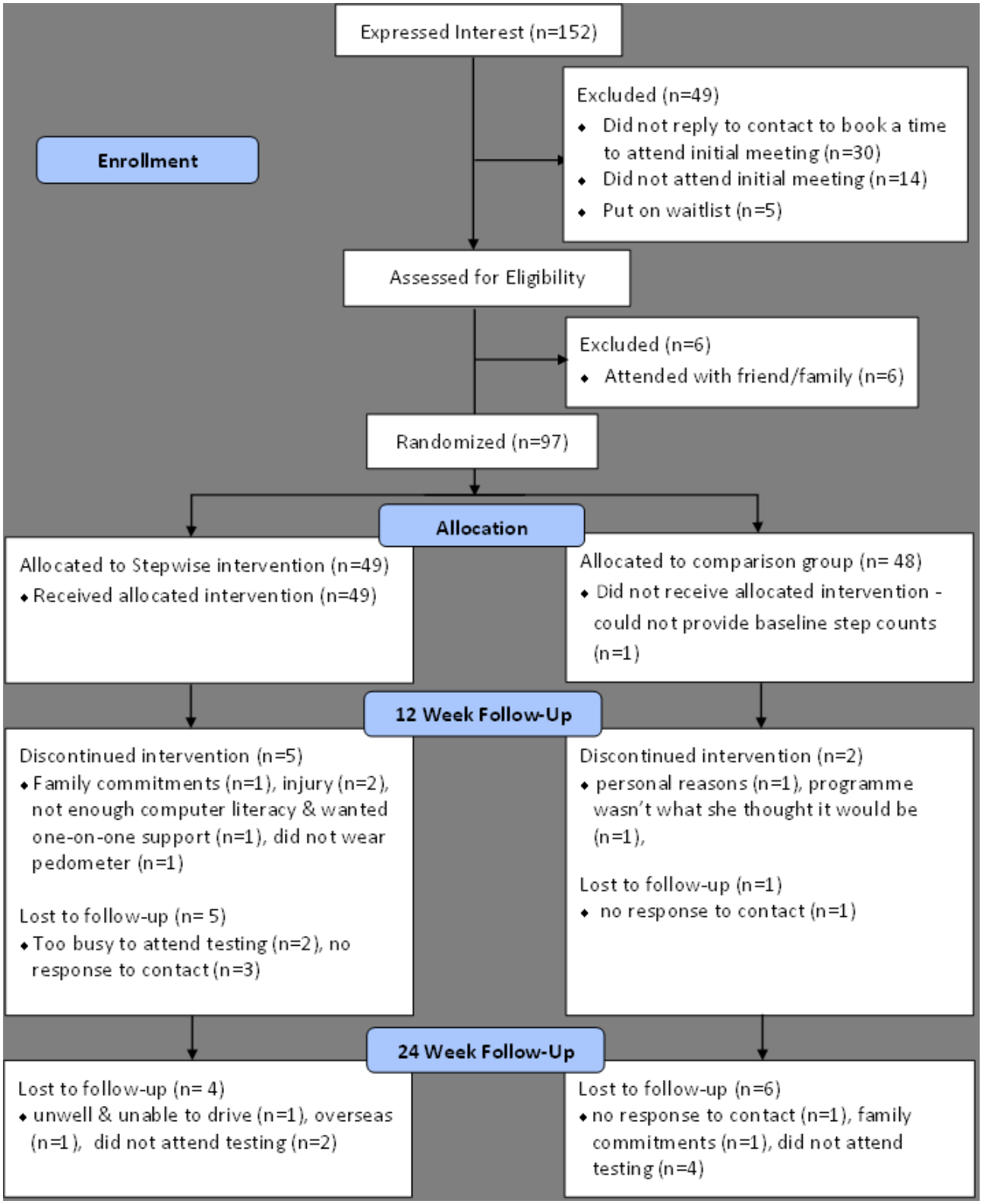
Baseline		StepWise (n=49) Mean (SD) or n (%)	Comparison (n=48) Mean (SD) or n (%)
Age (years)		47.1 (11.3)	45.3 (11.1)
Height (cm)		165.8 (7.9)	166.3 (7.8)
Weight (kg)		85.8 (20.2)	85.7 (19.9)
Ethnicity			
	NZ European	35 (71)	40 (85)
	Other European	4 (8)	2 (4)
	Maori	2 (4)	2 (4)
	Samoan	2 (4)	1 (2)
	Other	6 (12)	3 (6)
Employment			
	Worked full-time	25 (51)	29 (60)
	Worked part-time	10 (21)	10 (21)
	Students	4 (8)	3 (6)
	Homemakers	3 (6)	3 (6)
	Self-employed	3 (6)	1 (2)
	Retired	2 (4)	1 (2)
	Unemployed	2 (4)	—
	Did not report	—	1 (2)
Education			
	University degree	30 (61)	36 (75)
	Secondary school	16 (33)	12 (25)
	No secondary school	3 (6)	—
Body mass index		31.2 (6.6)	31.0 (6.5)
Step counts (steps per week)		50,971 (16,069)	53,480 (17,717)
Resting heart rate		66.4 (8.4)	67.5 (10.6)
Diastolic blood pressure (mmHg)[b]		76.6 (9.4)	76.1 (12.9)
Systolic blood pressure (mmHg)[b]		119.5 (15.8)	119.1 (14.9)
Glucose (mmol/L)[c]		5.2 (0.6)	5.4 (0.8)
Total cholesterol (mmol/L)[c]		4.7 (0.9)	4.9 (0.9)
HDL cholesterol (mmol/L)[c]		1.4 (0.4)	1.4 (0.4)
Triglycerides (mmol/L)[c]		1.2 (0.6)	1.3 (0.7)
Positive affect		3.5 (0.7)	3.3 (0.5)
Negative affect		1.5 (0.6)	1.6 (0.5)
Perceived health		71.5 (17.2)	67.3 (14.5)

[a]There were no significant differences between the groups for any of the variables at baseline.

[b]millimeter of mercury

[c]millimoles per litre

Figure 3. Flow of participants through the study.

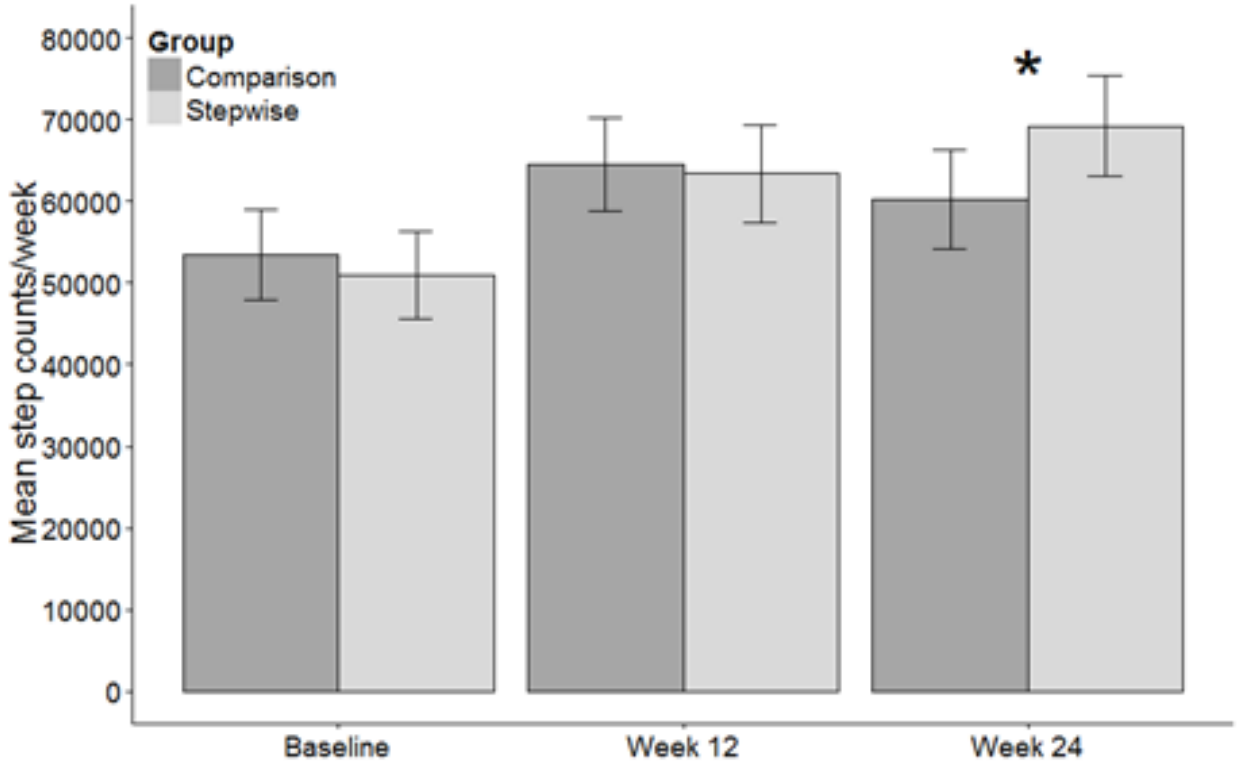


Primary Outcome: Step Counts

There were no group differences in step counts between baseline and 12 weeks. All participants increased their step counts per week from baseline taking 11,000 (CI 5739-16,266, *P*<.001) more steps at week 12 and 6,810 (CI 1190-12,431, *P*=.02) more at week 24 (see Table 2 for all significant effects of the intervention). However, an interaction effect showed the change in steps between week 12 and 24 was different between groups, SW took 8939 (CI 274-17,604, *P*=.04) more steps at week 24 than CP (see Figure 4). Separate comparisons for SW and CP showed that SW increased their step counts between week 12

and 24 (CI -11,826 to 112, *P*=.055). CP step counts did not change significantly (*P*=.15).

Figure 4. Mean steps per week taken by the SW and CP groups at baseline, week 12 and 24 (* denotes significant difference between groups, *P*=.04).



Secondary Outcomes

Health Related

There were no differences in any of the health-related variables between groups from baseline to week 12 or week 24. However, there were positive physical health changes for all participants across time. There was a mean reduction in triglycerides at week 12 (0.14 mmol/L, CI -0.25 to -0.02, *P*=.02) and decreased diastolic blood pressure (4.22 mmHg, CI -6.73 to -1.72, *P*=.003) at week 12 and week 24 (3.17 mmHg, CI -5.55 to -0.78, *P*=.03). Less positive was an increase in total cholesterol from baseline to week 12 (0.26 mmol/L, CI 0.099-0.423, *P*=.002) and 24 (0.38 mmol/L, CI 0.20-0.56, *P* = <.001) without a concomitant increase in HDL Cholesterol. There were positive effects for psychological health with increased positive affect and decreased negative affect at week 12 and participants perceived they were in better health at week 12 and 24 compared to baseline (see Table 2). There were no significant effects for fasting glucose, weight, resting heart rate or systolic blood pressure.

Table 2. Results of the step count and health data for the StepWise (SW) and Comparison (CP) groups at baseline, 12, and 24 weeks.

Outcome Variable	Group	Time			Significant Linear Mixed Model Effects				
		Baseline Mean (SE)	Week 12 Mean (SE)	Week 24 Mean (SE)		β	SE	95% CI	*P*
Weekly Step Counts	SW	50,971 (2747)	63,377 (3031)	69,229 (3176)	Time: Baseline to wk 12	11,002	2685	5739-16,266	<.001
	CP	53,480 (2804)	64,482 (2911)	60,290 (3081)	Time: Baseline to wk 24	6810	2868	1190-12,431	.02
					Time × Group Interaction: wk 12	1403	3895	-6231 to 9036	.72
					Time × Group Interaction: wk 24	11,447	4133	3346-19,548	.006
Total Cholesterol (mmol/L)	SW	4.67 (.12)	4.92 (.13)	4.89 (.14)	Time: Baseline to wk 12	.26	.08	0.10-0.42	.002
	CP	4.91 (.13)	5.17 (.13)	5.29 (.13)	Time: Baseline to wk 24	.38	.09	0.20-0.56	<.001
Triglycerides (mmol/L)	SW	1.21 (.09)	1.11 (.10)	1.10 (.10)	Time: Baseline to wk 12	-.14	.06	-0.25 to -0.02	.02
	CP	1.28 (.09)	1.15 (.10)	1.21 (.10)					
Diastolic blood pressure (mmHg)	SW	76.60 (1.35)	71.68 (2.06)	69.94 (2.62)	Time: Baseline to wk 12	-4.22	1.28	-6.73 to -1.72	.003
	CP	76.13 (1.91)	74.63 (1.90)	73.00 (1.50)	Time: Baseline to wk 24	-3.17	1.22	-5.55 to -0.78	.03
Positive Affect	SW	3.52 (.08)	3.65 (.09)	3.65 (.10)	Time: Baseline to wk 12	.21	.09	0.03-0.38	.02
	CP	3.33 (.08)	3.54 (.09)	3.36 (.09)					
Negative Affect	SW	1.53 (.07)	1.49 (.08)	1.48 (.08)	Time: Baseline to wk 12	-.15	.06	-0.28 to -0.03	.02
	CP	1.55 (.07)	1.39 (.08)	1.42 (.08)					
Perceived Health	SW	71.50 (2.13)	77.62 (2.34)	79.74 (2.43)	Time: Baseline to wk 12	6.37	2.15	2.10-10.65	.004
	CP	67.26 (2.18)	73.63 (2.26)	75.78 (2.36)	Time: Baseline to wk 24	8.52	2.29	3.98-13.06	<.001

Motivation

A time main effect showed that task self-efficacy ($F_{1.76,109.13}$=9.56, *P*<.001, η^2= .13) and autonomous motivation ($F_{2,128}$=10.12, *P*<.001, η^2= .14) increased over time while controlled motivation decreased ($F_{1.71,109.71}$= 5.80, *P*=.004, η^2= .08). Compared to baseline, participants felt more confident walking at week 12 (ΔM=9.58, CI 3.09-16.06, *P*=.002) and 24 (ΔM=9.24, CI 2.44-16.05, *P*=.004) had higher autonomous motivation at week 12 (ΔM=0.21, CI 0.06-0.36, *P*=.003,) and week 24 (ΔM=.25, CI 0.10-0.39, *P*<.001,) and had lower controlled motivation at week 24 (ΔM=0.21, CI 0.07-0.35, *P*=.001). There were no differences between the groups.

StepWise Website Usage

During the 12-week intervention all but 1 participant self-reported that they had accessed the StepWise website and 84% (28/33) stated they had used it at least once a week, with 15% (5/33) once every 2 weeks. The website usage statistics that were downloaded also confirmed this and showed that 82% of participants (27/33) logged into the website weekly, 1 participant logged in 8 out of 12 weeks, 4 logged in 9 out of the 12 weeks and 1 logged in 10 out of 12 weeks. Encouragingly, all participants used the step log function and therefore automatically received the goal accomplishment and tailored motivation messages and 45% (15/33) regularly used the activity plan function. Together, these results show the participants were engaged with the intervention and used the intervention tools provided on the website. At 24 weeks, self-report data and the website usage statistics showed that 48% of participants (16/33) were still accessing the website with 69% (11/16) accessing it (unprompted by email) at least once every 2 weeks.

Features of the SW Website Found to be Most Useful

Participants reported three website features they found most useful: (1) The step goals and pedometer because they provided a weekly challenge, (2) the self-monitoring tools of entering their step count each week and the graph to have a visual representation of the goal and to see whether or not it had been achieved, and (3) the feedback from the motivational messages that provided encouragement and positive reinforcement.

Discussion

Principal Results

We developed an intervention containing an individually tailored step goal pedometer walking program, combined with an evidence-based interactive website. In this pilot study we examined whether it would increase walking behavior, improve health and motivation over the short (12-week) and medium-term (24-week) in a community sample of apparently healthy adults. Results showed there were no differences in step counts between the SW and CP groups at week 12, all participants, irrespective of group, increased their step counts from baseline to week 12 and 24. However, importantly, SW participants had significantly higher step counts at week 24 compared to CP participants, suggesting that the intervention successfully helped individuals maintain their new levels of physical activity. In conjunction with the increase in walking, triglyceride levels and diastolic blood pressure improved as well as positive and negative affect (indicators of psychological well-being) and perceived health in participants of both groups. Surprisingly, total cholesterol increased across the study without a significant change in HDL cholesterol. Motivationally, all participants gained greater confidence for walking and their motivation became more autonomous, irrespective of their group assignment. These results are generalizable to a community sample of relatively healthy volunteers wanting to increase their walking behavior.

Participants increased their step counts by 21-24% above baseline at week 12. These results are smaller, but still comparable with, other pedometer interventions with similar samples that have shown increases of around 27% [13-15]. The CP group obtained the same increases in physical activity as the SW group over the short term and a number of components may have contributed to the increase. When participants are motivated enough to enroll in a walking study and know their activity will be monitored this can lead to short term behavior change [14]. Being provided with a pedometer and access to resources about increasing physical activity has also been shown to increase physical activity levels in the short term [51]. If the CP group had not been given a pedometer then, potentially, we would not have seen the same positive increases in physical activity. The strength of the SW intervention is that it resulted in maintenance of behavior change. At week 24 the SW group were achieving, on average, 1277 steps per day more (8939 steps per week) than the CP group and overall averaging 2608 steps per day more (18,256 steps per week) than their baseline values. The CP group achieved 973 steps per day more (6810 steps per week) at week 24 than baseline, and this was a decrease from the positive change of 1571 steps per day (11,002 steps per week) over baseline that they achieved at week 12. Therefore, as expected, the resources provided as the test of standard practice were not enough to maintain behavior change. Only the SW group achieved the goal of the walking program which was to increase their weekly step counts by 15,000 above baseline and meet the physical activity guidelines for health [31]. This is important because few Web-based walking interventions are able to demonstrate (or have not measured) sustained changes in physical activity resulting in the achievement of physical activity recommendations. For example, Carr et al [34] reported their participants' step counts had returned to near baseline levels at 8 months following their intervention. While a similar study to ours, Carr et al [52] reported that their enhanced Internet group had higher physical activity levels (measured by 7-day physical activity recall) at 12 weeks compared to their standard Internet group (publicly available physical activity websites) but that there were no differences between the groups at 24 weeks. Ideally, future research should continue to monitor behavior change for longer than 24 weeks.

Consequently, despite the lack of group differences at 12 weeks, providing participants with a step goal walking program and access to a website containing the evidence-based behavior change tools to self-monitor their behavior, get feedback and motivational support, and plan their activities, seemed to encourage individuals to maintain the proposed increased physical activity at 24 weeks [6,11-12]. The success of the study was dependent on participants using the website. Encouragingly, both our objective and self-report data showed that 97% engaged the website during the intervention period at least once every 2 weeks. During the follow-up period, when there were no email prompts sent to remind participants to access the website, 48% of participants were still accessing it. These statistics suggest we achieved the goal of creating a website that individuals continued to use. This overcomes one of the reported limitations of Web-based studies, that visits to the website (and therefore exposure and engagement with the intervention) decrease over time [12,37]. Arguably, the content of the website is the key to helping individuals initiate and maintain behavior change. The ability to record steps and see goal achievement was stated as one of the most important features of the website and has been shown previously to be important for successful physical activity behavior change [22-23,52-53]. Alongside the changes in physical activity, there were significant health-related improvements. The decreases in diastolic blood pressure (4.2 mmHg at week 12 and 3.2 mmHg at week 24) were similar to those found with other walking interventions [7,54] and of clinical significance. A 2 mmHg reduction is estimated to reduce the incidence of coronary heart disease by 6% and of stroke by 15% [55]. Our intervention resulted in 0.1% decrease in triglycerides from baseline to week 12. Although this is a modest decrease, a larger effect would not have been expected since our sample had normal levels of triglycerides at baseline [56]. The extent of the decrease in triglycerides required to benefit health remains unclear [57]. The increase in total cholesterol was unexpected, particularly since HDL cholesterol did not change, however, the average values still remain in the normal range [56]. More positively, there were significant improvements in psychological well-being with individuals reporting greater

levels of positive affect and reduced negative effect as well as improved perceived health. The change in positive affect and perceived health was also shown by Fitzsimons et al [33] who employed the same step-goal-based walking program but in a non-Web-based setting.

The study was successful in changing motivation for physical activity in both groups. Participants felt more confident in their ability to walk for longer periods of time. Increased confidence has been demonstrated in a number of walking studies and is a strong predictor of physical activity behavior [58]. The personal performance successes participants could see from their step counts increasing (both groups) and achieving goals (SW group) would have contributed to the increased confidence [58-59]. Participants had greater autonomous motivation at the conclusion of the study compared to the baseline, meaning that they were more motivated to participate in physical activity out of enjoyment and for the value they attached to the outcomes of being active. Furthermore, they had less controlled motivation at the end of the study meaning they were not feeling as pressured to participate in activity. Together these changes mean individuals are more likely to *want* to be physically active rather than feeling they *have* to be physically active and the changes are related to increased and sustained physical activity behavior [60]. Interestingly, the change in controlled motivation did not occur until week 24 when participants had completed the intervention. It may be that the act of being involved in a pedometer-based study which required self-monitoring of behavior (SW) or prompted individuals to decide to self-monitor (CP) can act as a controlling pressure on behavior. The impact of pedometers on autonomous and controlled motivation has not been investigated and would be an interesting avenue for future research, to ensure pedometer-based interventions do not have a negative effect on motivational quality.

Limitations

Both groups received a pedometer and physical activity device, therefore the study did not have a no treatment control group. Consequently, there is still the possibility that some other factor other than the elements contained in the intervention and comparison group caused the change in physical activity behavior. Step counts were only recorded for analysis at baseline, 12 weeks and 24 weeks. It may be that participants simply increased their walking behavior in the measurement week and these values do not actually reflect the activity they achieved during the intervention. However, for someone to have such a large increase in activity in one week is unlikely. The walking program automatically increased weekly step count goals, so consequently if something happened to disrupt (decrease) normal walking patterns (eg, getting ill) then the individual would be expected to meet a new, harder goal on resumption of the program. Ideally, the person would have been prompted by the program to go back to the walking goal from the week prior to the disruption. However, our StepWise software was not able to do this. This should be taken into account in future development of the intervention. Additionally, the intensity at which participants walked was not measured. Although we endeavored to emphasize that participants walked at moderate intensity, we cannot be sure they adhered to this. It was hoped that the study would have 80% power to detect differences in step counts (the primary outcome), however, after drop-outs we did not achieve our target sample size of 43 per group. Our large drop-out rate is a limitation; we were able to elicit reasons for why 9 SW and 3 CP participants decided not to continue with the study but we were not able to ascertain the reasons why 5 SW and 6 CP did not show to the testing sessions. As a result, we cannot be sure whether there was some underlying factor that caused these individuals to drop out and therefore bias the results. Furthermore, it is likely the study was underpowered to detect differences in the biochemical health measures taken. Finally, in hindsight, it would have proved useful to also ask participants if there were any other features they would have liked the website to have to encourage and support their physical activity. This would have been advantageous for future development of the StepWise website.

Conclusions

There were no differences in step counts between the SW and CP groups following the 12-week intervention; all participants increased their walking behavior and had improved physical and psychological health. However, only the StepWise group successfully maintained their increased walking behavior 3 months post-intervention and achieved step counts that met the physical activity guidelines for health. Sustained behavior change is supported by a step-goal-based walking program with behavior change tools that allow a person to self-monitor behavior, get feedback and motivational support, and make activity and coping plans.

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Authors' Contributions

EAH was the principal investigator and along with NM conceived the idea for the study and developed the methodology. EAH created the content for the StepWise website, oversaw the data collection, conducted the analysis and drafted the manuscript. NM critically reviewed the manuscript drafts. JDF contributed to the content of the StepWise website, coordinated the data collection, prepared the data for analysis and reviewed the manuscript drafts.

Conflicts of Interest

None declared.

Multimedia Appendix 1

CONSORT EHEALTH form V1.6.

[PDF File (Adobe PDF File), 15MB - resprot_v5i1e14_app1.pdf]

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Abbreviations

BCT: behavior change technique
CP: comparison group
HDL: high density lipoprotein
SW: StepWise intervention group

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Protocol

Digital Gaming for Improving the Functioning of People With Traumatic Brain Injury: Protocol of a Feasibility Study

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Abstract

Background: Traumatic brain injury (TBI) is a critical public health problem. The recovery process for people with TBI is typically slow and dependent on complex and intensive assisted rehabilitation programs.

Objective: To evaluate the effects and feasibility of digital games for cognitive functioning and general well-being among people with traumatic brain injury.

Methods: This is a single-site feasibility study conducted in Finland, which uses a pragmatic, randomized controlled trial with three arms, and will recruit patients from the Turku University Hospital, Division of Clinical Neurosciences in Finland. Participants must meet the following inclusion criteria: (1) a Finnish speaking adult, aged 18-65 years; (2) diagnosed with a traumatic brain injury (diagnostic criteria ICD-10, S06.X, T90.5) in the University Hospital; (3) access to a TV, a computer, and the Internet at home; (4) not an active digital gamer (5 hours or less a week); (5) willing to participate in the study. Participants must have been discharged from the neurologic treatment period for traumatic brain injury for over 12 months before the commencement of the trial, and they may not have actively participated in cognitive rehabilitation during the 3 months prior to the trial. Written informed consent will be mandatory for acceptance into the trial. Exclusion criteria are as follows: (1) sensory, cognitive, or physical impairment (eg, severe cognitive impairment); (2) a deficiency restricting the use of computers or computer game control system unaided (eg, impairment in vision, severe astigmatism, hemiplegia, disorder in visuospatial perception, dysfunction of the central vestibular system); (3) apathy identified in previous neuropsychological evaluations; (4) diagnosed severe mental disorders (eg, schizophrenia or severe depressive disorders to be identified in medical records as the secondary diagnosis).

Results: The preparatory phase for the study is fulfilled. Recruitment started in June 2015 and finished November 2015. Results will be reported in 2016.

Conclusions: The specific outcomes such as primary outcome measures were selected because they are widely used psychological tests and thought to be sensitive to changes in the cognitive functions related to TBI.

Trial Registration: Clinicaltrials.gov NCT02425527; https://clinicaltrials.gov/ct2/show/NCT02425527 (Archived by WebCite at http://www.webcitation.org/6esKI1uDH)

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KEYWORDS

digital games; brain injury; cognitive rehabilitation

Introduction

Traumatic brain injury (TBI) is a critical public health problem. In the USA alone at least 5.3 million citizens live with disabilities resulting from TBI [1]. In Finland there are about 40,000 people with TBI [2] and approximately 100,000 people live with disabilities resulting from brain injuries [3]. The most common groups with brain injuries are teenagers and young adults between the ages of 15 and 34. Treatment of TBI is long lasting with long-term care, which therefore incurs economic cost to health systems [4]. Impairments, apathy, mood disorders, impulsivity, and changes in personal character [3] or deficits of attention and short-term memory are among the most common and disabling characteristics of people with TBI [5]. The recovery process is typically slow and depends on complex and intensive assisted rehabilitation programs [6-8].

Although physicians have long believed that individuals with brain injury benefit from early and long-standing therapeutic intervention [9], direct evidence is still scarce [10]. A recent expert panel on cognitive rehabilitation suggested that metacognitive strategy training focusing on functional everyday activities is appropriate for people with TBI [11]. A systematic review by Spreij et al suggests that computer-based cognitive retraining is considered to be the most promising novel approach of the last decade, because of the positive results in improving memory function after an acquired brain injury [12]. Training with video games is also a rapidly developing area for industry, since it has been estimated that the average young person has played a total of 10,000 hours of games by the age of 21 [13]. Virtual reality games have been found to improve patients' mobility [14] or motor learning [15]. A meta-analytic study by Toril et al [16] indicated based on 21 experimental studies and 474 trained and 439 healthy older controls that video game training produces positive effects on cognitive functions, including reaction time, attention, memory, and global cognition. However, inferences from the results have to be made cautiously because of the problem of the high heterogeneity of the studies. Further, a systematic review concluded that video games have the potential to improve health outcomes in psychological and physical therapy [17]. On the other hand, Lampit and colleagues [18] found that computerized cognitive training in healthy adults is modestly effective at improving cognitive performance, but efficacy varies across cognitive domains and is largely determined by design choices.

Some encouraging results for virtual reality interventions have been observed in patients with post-stroke conditions [19]. For example, those who are active players show better performance in alertness and cognition compared to those who do not play games [20,21]. In addition, the game-based exercises in recovery process have shown increased motivation to perform rehabilitative tasks [22]. Further, preliminary results have shown that playing action video games has produced significant improvements in attentional control for healthy adults [23].

Still the scientific evidence for this area is at best mixed. Ball and colleagues [24] conducted a large-scale cognitive training study and found that when memory, attention, and problem solving were trained independently, trainees improved each skill respectively. However, there was no transfer to other untrained skills. Game programs were also found to be inadequate for efficient integration in current clinical practice [25,26]. Stand-alone systems without any human interaction have also been criticized [27]. In addition, there is still a lack of information regarding what kinds of games participants want to play as well as the effectiveness of serious games for patients with brain injury. Gravel and colleagues [28] concluded in their systematic review that there is a paucity of well-designed clinical studies for patients who sustain mild TBI. The large variability in outcomes measured may also limit comparison between studies. Although experiments using games among people with brain injury have been conducted, the effectiveness of commercially available games has been evaluated to a lesser extent [19]. As traumatic brain injury causes long-term disability with adverse social, psychological and economic consequences, it is therefore important to seek methods to optimize independence and social participation to reduce long-term care needs and enhance quality of life among people with traumatic brain injury [29]. The meta-analyses by Lohse and colleagues [19] also indicate that more evidence is needed to evaluate the effectiveness of games among people with brain injury.

The aim of this study is to evaluate the effects and feasibility of digital games for cognitive functioning and general well-being among people with traumatic brain injury.

Methods

Trial design

This is a single-site feasibility study, conducted in Finland, which uses a pragmatic, user-centered, randomized controlled, three-arm parallel-group design.

Participants

Patients will be individually and randomly assigned to one of the three parallel groups, initially in a 1:1:1 ratio, to an intervention group (rehabilitation gaming), active control group (entertainment gaming) or passive control group (no gaming) organized by the research study.

To be eligible for this study, participants must be Finnish speaking and reading adults between the ages of 18 and 65, who meet the eligibility criteria for people with traumatic brain injury (diagnostic criteria ICD-10, S06.X, T90.5) Participants must

have been discharged from the neurologic treatment period due to traumatic brain injury over 12 months before the recruitment and must not have been actively participating in cognitive rehabilitation during the last 3 months. They must own a TV, a computer and have internet access at home, but must not be active digital gamers (5 hours or less a week). Potential participants will not be admitted to the study if they have any of the following exclusion criteria: sensory (eg, serious visual impairment), cognitive (eg, memory problems, slow processing speed, a lack of attention, linguistic problems) or physical impairment (eg, severe cognitive impairment or impairment in vision, severe astigmatism, hemiplegia, disorder in visuospatial perception, dysfunction of the central vestibular system), which may restrict the use of computers or computer game control system unaided; apathy identified in previous neuropsychological evaluations; a diagnosis of a severe mental disorder (eg, schizophrenia or severe depressive disorders to be identified as the secondary diagnosis); or having been active in cognitive rehabilitation during the previous 3 months. The primary source to assess patient eligibility will be the university hospital electronic medical records and later telephone calls and interviews with a trained psychologist.

The study will take place at the Turku University Hospital, Division of Clinical Neurosciences (Operational Division of Clinical Neurosciences, Department of Rehabilitation and Brain Trauma, in collaboration with former neurologic outpatient clinic and rehabilitation units of the university hospital) located in southern Finland. The Division is specialized in neurological and psychiatric inpatient rehabilitation, inter-professional neurological education, rehabilitative examinations, and the inpatient and outpatient care of traumatic brain injuries. Each year, about 350 patients are admitted to the Division services units [30].

The randomization (started in June, 2015) will target patients who have been registered in the Turku University Hospital electronic medical records. The recruitment will focus on patients who have been discharged from the Turku University Hospital, Division of Clinical Neurosciences at least 12 months before the recruitment. It has been indicated that using games in longer interventions will be facilitated more easily if training is provided in the patient's own environment, such as home, due to easier access and reduced impact on school or work activities [31]. To capture a representative sample of patients, the Turku University Hospital electronic medical records will be accessed by the authority of the chief medical doctor of the Hospital together with the Research Assistant. All patients with a TBI diagnosis will be screened to determine which patients fulfill the eligibility criteria for study participation.

Interventions

Patients will be randomly assigned to rehabilitation gaming (intervention group), entertainment gaming (active control group), or passive control group (no gaming).

First, the rehabilitation gaming (intervention group) will use an internet browser-based digital brain training program, CogniFit [32]. This web-based cognitive training platform includes about 33 games designed with the purpose of improving the user's cognitive abilities as brain exercises. In this study, we aimed at controlling for the type of "cognitive demands" the games pose to the player by preselecting games that share certain features while allowing participants to choose a game that they find pleasurable to engage with. For the purpose of the research, a selection of games will be included in the intervention. The game tasks target different cognitive abilities such as memory, visual-spatial ability, attention, tracking and fast decision making. The intervention in our study is user-centered. This means that participants are instructed to play at least one exercise from each of the four categories during each training session daily; they are otherwise free to choose which exercises they wish to play. It is reasonable to assume that in real life people would choose to play games that they like. The possibility to choose a game that you like is supposed to increase the likelihood that participants engage in gaming as instructed. The game tasks can also be adapted to players' skills getting more difficult as the players progress. CogniFit requires a computer with Internet access.

The participants will have their own user account on CogniFit, which will record the games they have selected, how often and how long their playing session has taken, and how they have progressed in each game. Access into CogniFit will be introduced to the participants during the introductory meeting by the Research Assistant. To assess the amount of time Research Assistants are spending with each participant, the length of the introductory meeting will be recorded. This will ensure that we are aware of the level and time of support offered to each participant before the intervention start. Any help later at home or by telephone will be offered by a Technical Assistant if needed; any contacts with the Technical Assistant are recorded. The participants will be guided to use the rehabilitation gaming for at least 30 minutes per day over a period of 8 weeks. Daily gaming sessions are supported by studies [13,33,34], although opposite suggestions have also been found in the literature. For example, the meta-analytic study by Lampit and colleagues [18] concluded that training more than 3 times per week in an unsupervised home environment was specifically ineffective for healthy older adults. To encourage, motivate, and hold participants to training in each category, they will be supported in planning a schedule for their training sessions (days, time, and frequency) by the Research Assistants for the entire 8 week gaming period. The participants will also be encouraged to seek help and support by telephone. If requested by the participant, a technical assistant will visit the participant's home to help set up the computer in a quiet place and offer training [35].

Further, to support the participants' gaming activities, their ability and previous experience in playing digital games will be explored. This will ensure that the participants have basic gaming skills required for active gaming. A description of the overview of the program will be offered. A new email address, the passwords for the email account and personal account will be generated. This is necessary because the browser-based program requires access through a website and the user will log in with an email address and a specified password. The participant will also experiment with the game unaided to find out possible barriers in his/her gaming. The program will record participants' progression and scores on each of the games. In

order to do this, the research team will have access to the program to monitor the progress of each participant's game scores. Information about the frequency of training sessions will also be recorded by the participants themselves into a specific calendar. Based on the information in the diary, we can statistically control for the effect of actual amount of gaming on the results. Beside the participants' diary, participants' adherence to and motivation [36] for gaming will be supported and monitored by weekly telephone calls by the research assistants.

Second, the active control group, (ie, "entertainment gaming") will use commercial digital games designed for Sony Playstation 3 (PS3) consoles. PS3 consoles will be connected to the participant's TV systems via a HDMI cable or an RCA connector cable; using a console does not require an internet connection. The games will be played with wireless and rechargeable Sony DualShock gamepad controllers. As in the intervention group, the participants chose a game that they found pleasurable to engage with. The possibility to choose a game may increase the likelihood that participants engage in gaming as instructed. Participants will have already selected the most favored game at home out of eight possible games. Entertaining games were carefully selected to correspond to the CogniFit games. Although the eight games differ from each other with respect to aesthetics and narrative qualities, each game contain the same core gameplay. Gameplay can be defined as consisting of two elements: the challenges the designed game presents for the player as the game proceeds and the actions the player decides to take to overcome the challenges and to reach in-game goals [37]. The core gameplay of each of the eight games consist of: 1) real-time action which demands quick responses and decisions; 2) identifying relevant information from irrelevant noise; 3) targeting, tracking and shooting; 4) orienting and navigation in 3D space; and 5) tasks that require memorizing and reasoning.

The attractiveness of the game for the player will also be ensured. This, in turn, is thought to increase the motivation to engage in training. We thought that it is important that participants are not forced to play, for example, violent games if they prefer not to. It is reasonable to assume that in real life, people would choose to play games that they like.

The game selected will be played together with the Research Assistant during the introductory meeting. To assess the amount of time Research Assistants are spending with each participant, and the needs depending on the type of console, the length of the introductory meeting will be recorded. This will ensure that we are aware of the level and time of support offered to each participant. As with the intervention group, ability to play digital games will be explored to ensure that participants have the basic gaming skills required for active gaming. An overview of the use of the PS3 console will also be offered and a tutorial demonstration will be given. This will include, for example, how to start the console, how to play the game, how to use the controller, how to change game options (ie, game difficulty and speed). The participant will be able to try the game that they have preselected from the list of eight games. In addition, the project will purchase the game for the participant either by buying the game from 1) the official Playstation Store and downloading and installing the game into a new PS3 console that will be offered to the participant for intervention use at home or 2) a retail store. If requested by the participant, a Technical Assistant will visit the participant's home to help set up the console in a quiet place and offer training [35] or guidance by telephone. As with the intervention group, the participants will be guided to play the console for at least 30 minutes per day over a period of 8 weeks [13]. To assess the amount of time Research Assistants are spending with each participant, the length of the introductory meeting will be recorded. This will ensure that we are aware the level and time of support is offered to each participant before the intervention start. Any help later at home or by telephone will be offered by the Technical Assistant if needed; any contacts with the Technical Assistant are recorded. To encourage, motivate, and hold participants to a training schedule, participants will be supported in planning their training session schedule (days and times) with the Research Assistant for the entire 8 week gaming period. Information about game sessions (day, time, frequency, play progress) will also be recorded by the participant into a calendar (gaming diary). Based on the information in the diary, we can statistically control for the effect of actual amount of gaming on the results. Further, adherence to gaming will be supported and monitored by weekly telephone calls by the Research Assistant. The participants will also be able to change the game during the 8-week period if they have concerns due to violent content, for example.

Third, the participants in the passive control group will not have gaming activities organized by the project. As with the intervention groups, the Research Assistant will call weekly to the participants in this study arm. In addition, as patients in a control group, these participants will be offered an opportunity to have games and consoles for a 2 week period free of charge after the study. Further, 5 out of the 90 participants will have a chance to randomly win a console to keep after the study intervention period.

The procedure for patient recruitment and allocation will consist of specific steps. First, potentially eligible participants will be selected from the hospital patient records based on the inclusion criteria with the help of a specialist in neurology. Those patients who have been screened and assessed a prior to meet the inclusion criteria will be contacted by telephone by the Research Assistant. Any cognitive and physical problems limiting patients' ability to participate in the study will be discussed by phone. If a patient cannot be reached by the telephone call, a written information letter about the study will be posted to him or her. If interested, he or she can contact to the Research Assistant. A preliminary description of the study will be offered, and the inclusion criteria for the study will be described to the potential participants. Based on the discussion with the participants, their eligibility for the study will be decided. Eligible participants will also be asked about preliminary interest in the study. They will be informed that they will receive information about the study by post, 2 informed consent forms, questionnaires to be filled in (baseline data), and a short description of the 8 optional games (in case of allocation to the control group).

Possible participants will also be made aware that after 1-2 weeks they will receive another call from the Research Assistant. This will ensure that potential participants have enough time to read all materials, ask questions, make the decision to participate in the study or not and decide what type of game they would like to play if allocated to the group of entertainment gaming. During this telephone call, those who are willing to participate in the study will get information about the practical arrangements of the study and the place of the data collection. They will also be informed that all travel costs will be covered and that there will be a face-to-face meeting scheduled that may take 1-1.5 hours in total. In addition, the participants will be made aware that they are free to withdraw from the study at any time.

During the face-to-face meeting at the research laboratory, participants will be confirmed as successfully or unsuccessfully meeting the inclusion criteria. The current cognitive status of the participant will be initially assessed during the face-to-face meeting with the Research Assistant. After the initial interview, a battery of neuropsychological tests will be used in the baseline measurement phase (see primary and secondary outcome measures). If during testing it is obvious that the participant suffers from severe cognitive impairments that prevent the use of regular computers or games, the participant will be excluded from the study.

Eligible participants will sign an informed consent form of their own free will. Should a person consent, their previously-collected baseline data will be gathered. Cognitive measurements will be then conducted by a trained psychologist. After that, the trial manager will receive a message by email, text message or telephone about each eligible patient and will allocate the patient to one of the three arms of the trial, based on a computer generated randomization list (intervention group vs. active control group vs. passive control group). The Research Assistant will receive this information. Recruitment will continue until all required data has been received.

Results

The preparatory phase for the study is fulfilled. Recruitment started in June, 2015 and finished September, 2015. Results will be reported in 2016.

Discussion

Outcomes

The specific outcomes, such as primary outcome measures (The Trail Making Test, parts of the Wechsler Adult Intelligence Scale, 4th Edition, or WAIS-IV) were selected because they are widely used psychological tests [39,42]. They were thought to be sensitive to changes in the cognitive functions typically related to TBI. Moreover, these functions were assumed to be trained by the video games. The Trail Making Test is one of the most used neuropsychological tests globally, allowing comparisons to previous studies on TBI and rehabilitation. WAIS-IV tasks also are among the most used psychological tests in the world, and they are sensitive enough to discriminate even healthy adults. WAIS-IV has been standardized also in Finland - the home country of the participants - allowing us to compare our sample to national standards. The secondary outcome measures were selected so that a comprehensive picture of the potential changes in cognitive functions that can be assumed to be trained by the games (both the video games and the cognitive training game) is obtained. Moreover, we thought it is important to record changes in mood and self-efficacy as these influence daily life of a patient. Finally, for practical purposes it is important to gain knowledge of the feasibility of the games, as measured by adherence to the treatment and the participants' personal experience with the game.

Primary Outcome

Processing Speed and Visuomotor Tasks

- *The Trail Making Test (TMT)* requires visual search, scanning, speed of processing, mental flexibility, and executive functions [39]. The test consists of two parts. In the first part, patients are given a paper displaying circles numbered 1 to 5 in random order. The patient is told to draw lines that will connect the numbers in ascending order. In the second part, there are circles with numbers from 1 to 13 and letters from A to L. This time, the patient draws lines to connect the circles so that they alter between numbers and letters in an ascending order (for example: 1-A-2-B-3-C) and so on. The time it takes to complete the trail is measured in both parts of the test. Errors do not affect the final score, but they must be corrected by the patient [40,41].
- *WAIS-IV tasks* (symbol search, coding and cancellation) are a test package used to measure cognitive skills of adult patients, especially skills of sorting out simple visual information, monitoring, making progress in the task, maintaining attention, visuomotor co-ordination and visual memory [42]. The tasks constitute 4 indices that measure different areas of cognitive skills. In symbol search and cancellation tasks, the participants perform a visual search in order to find out if a certain symbol is in the midst of other symbols. In the symbol search task, the symbols are organized in rows, and the participant has to indicate whether or not the required symbol was on each row. In the cancellation task, the participant looks for the same symbols during the whole task. In the coding task, the participant is given a set of numbers and symbols that match each other. Their task is to fill out an empty grid containing only numbers with the appropriate symbols matching those numbers. The test-retest reliabilities of the tasks used in this study ranged from 0.78 to 0.86 over an 8-82 day period in the original WAIS-IV standardization study [42]. A version of the test [42] containing norms for the Finnish population is used in this study.

Secondary Outcomes

Attention and Executive Functions

A *Simon task* [43,44] will be used to measure the inhibition component of executive functions [45]. This is a computerized visuomotor task in which correct responses and reaction times are measured. In the task, a blue or red square appears on either the left or right side of the screen. The participant is instructed

to push the left button on a response pad each time a blue square appears and the right button each time a red square appears, irrespective of which side the square is presented. In congruent trials, the response button is on the same side as the square, and in incongruent trials the square is on the opposite side of the response button (ie, the irrelevant spatial information is conflicting with the correct response). The differences in reaction times and error rates between the incongruent and congruent trials (the Simon effect) are used as the dependent measurements in this task. These variables reflect the extra processing cost of having to inhibit the incompatible spatial location of the stimulus.

Working Memory

- *WAIS-IV* (digit span) measures working memory [42]. In the first part of the task, the participants repeat numbers in the order they hear them. This task measures memory, coding, attention and auditory processing. In the second part, they repeat the numbers backwards. This task requires the use of working memory, processing information before giving an answer, internal processing and visuospatial organization. In the third part of the task, the participants repeat numbers in numerical order. This task again measures working memory and internal organization of the stimulus, but is more demanding than the other two tasks [42].
- *The Paced Auditory Serial Addition Test* (PASAT) was originally developed by Gronwall [46] to measure auditory information processing speed, flexibility and calculation skills, in order to monitor the rehabilitation of patients with mild head injuries. This study includes a well-established version of the test in which the stimulus presentation rates were adapted for MS-patients by Rao and colleagues [47]. In the first part, single numbers are presented every 3 seconds. The patient adds each new number to the last number prior to it. In the second part, the numbers are presented every 2 seconds, but the task is the same. The test score is the number of correct sums given in each trial.

Depression

The Patient Health Questionnaire (PHQ-9) [48,49] includes 9 items where respondents are asked to indicate with four-point scale how often they have been bothered by any of the problems over the last two weeks (0 = not at all; 1 = several days; 2 = more than half of the days; 3 = nearly every day). Based on the individual items, a total score is formed: the higher the score, the more severe the depression symptoms (range 0-27).

Self-Efficacy

The General Self-efficacy Scale (GSC) [49] has been created to assess a general sense of perceived self-efficacy to predict coping with daily hassles as well as adaptation after experiencing a variety of stressful life events. The scale for 10 items is self-administered and responses are made on a 4-point scale (1 = Not at all true; 2 = Hardly true; 3 = Moderately true; 4 = Exactly true). It takes about 4 minutes to complete. The final composite score ranges from 10 to 40, and comprises the sum of all 10 responses: low scores represent a lower ability to cope with daily problems.

Executive Functions

The Behavior Rating Inventory of Executive Function-Adult Version (BRIEF-A) is a 75-item questionnaire that focuses on executive functions in daily life [50]. The questionnaire consists of a behavioral regulation index (inhibition, shifting, emotional control and self-monitoring) and a metacognition index (initiation, working memory, planning/organizing, task monitoring and of materials). The answers to this self-administered questionnaire are given in a three-point Likert scale format (never/sometimes/often). A global executive composite (GEC) will be formed by the total score. The test has been found to be reliable (an internal consistency of α .80-.94 for the clinical scales; α .96-.98 for the indexes) [50].

Feasibility

- *Adherence:* Willingness to participate in the study (participation/refusal, yes, no); dropout for any reason (yes/no); involvement in the interventions for 8 weeks period (yes/no)
- *Usability:* "Was the game usable?" (yes/no)
- *Satisfaction:* "Have you been satisfied with the game?" (yes/no)
- *Use:* "Would you like to use the game in the future?" (yes/no/maybe)

Background information including socio-demographic characteristics and illness history will be collected (age, gender, marital status, level of education, employment status, living situation, illness history, illness history, current digital game playing (hours a week). If needed, detailed information about each participant's diagnosis and the time of traumatic brain injury will be collected based on electronic medical records to limit the amount of information to be collected from patients themselves.

Time for the Data Collection and Follow-Up

Patient data will be collected at three different times: at baseline, after the intervention (8 weeks), and as a follow-up, 3 months after the intervention. If required by the participant, he or she will receive a text message about 2 days before the meeting to remind them of the coming follow-up meeting.

Sample Size

The calculations for the sample size needed in each group are preliminary estimations to guide our data collection, which are based on previous studies: (1) TMT (version A) and (2) depression (PHQ-9). First, if a score on the TMT version will be about 71, the mean change in the scores during the follow-up will be 30, and standard deviation of the TMT scores will be 53 [41]. This difference between groups could be expected to be significant (with a power of 85%, p=.05) if the sample size in each group is 30 subjects. Second, if the average level of the PHQ-9 score is about 10, the mean change in the scores during the follow-up is then 3 (SD 5) [48]. The difference could be expected to be significant (with a power of 85%, p=.05) if the sample size in each group is 27 subjects. Thus, based on these preliminary power calculations, the sample size to be used in this study (30 in each group) is not very strong but will be reasonable for a feasibility study aiming to detect preliminary changes within the group, between baseline and follow-up

outcome measurements. However, this means that the attrition rate of the study should be near 0%. The outcomes of this study will be used as a guide to estimate a sample size for a multi-center study to be conducted, hopefully in the future.

Randomization

The study will be individually randomized. The randomization and patient allocation will be fully centralized. A central randomization service provided by the University of Turku will be used. Allocation will be computer generated. The participants will be randomly assigned (a block randomization in three blocks) using numbers via computerized assignment developed by the independent trial statistician and implemented by the Research Assistants (2), who are trained in patient randomization and data collection.

The research assistants overseeing patient recruitment and randomization will be aware of the assignments. Allocation will not be masked to participants in intervention and control groups or Research Assistants who will recruit patients. The psychologists, as cognitive outcome assessors, will be masked. The data analyst (the trial statistician) will be kept blinded to the allocation. As far as we are aware, there will be no contact between participants in different groups; participants will come from a wide geographic area in the catchment area of the Southwest Hospital District in Finland.

Analysis Plan

All the participants will be analyzed at the baseline, after 8 weeks when the intervention has been finalized, and at 3 months, using all the scales for primary and secondary outcomes. Comparisons in possible differences between means in 3 intervention groups and 3 time points will be conducted using analysis of variance (ANOVA test). Possible post hoc tests may be performed using paired *t*-tests, or Chi square tests to detect significant differences before and after treatment in sub-groups, if needed. Further, significant differences between groups will be evaluated applying the unpaired Student's *t*-test using the conventional 95% level of confidence. Statistical analyses will be done using SAS System for Windows, version 9.4 (SAS Institute Inc.). *P* values less than 0.05 will be considered statistically significant.

The cumulative monitoring data during the 8-week period regarding gaming for the total number of received parameters (frequency, timing, time) will be calculated. Feasibility information (secondary outcomes) available for each patient will be calculated (participation/refusal rate, measurement instrument filled, drop-outs, acceptability, usability, satisfaction, willingness) and compared to the highest possible representative numbers (100% participation rate; measurement instrument filled, drop-out rate 0%, acceptability 100%, usability 100%, satisfaction 100%, willingness to use games in the future 100%).

Criteria for feasibility are as follows:

- Willingness to participate 80% (refusal rate 20%)
- We expect that more than 75% of the prescheduled measurements will be performed (adherence).
- Less than 25% of participants drop out for to any reason (adherence)
- The acceptability of the game use is at least 95% (in intervention and control groups) (adherence).
- Usability evaluation for the gaming system is at least 80% (1 item; dichotomous scale yes/no) (usability).
- More than 80% are satisfied with the games (1 item; dichotomous scale, yes/no) (satisfaction)
- We expect that 60% are willing to use the games later as part of their recovery process (use in the future).

Ethical Issues

A proposal for this study has been evaluated by the ethical committee of the Turku University Hospital (ETMK 41/1801/2015). The permission to conduct the study has been granted by the Turku University Hospital (T89/T04/008/2015). The trial has been officially registered (NCT02425527). All patients will be informed of the study orally and in written format. Informed consent will be obtained for experimentation with human subjects. The neurologist is a part of the study group and an expert in this kind of treatment. Study participation is completely voluntary (a signed informed consent, the Declaration of Helsinki 2013).

Possible ethical issues have already been taken into account in the development phase of the study: the rehabilitation and entertainment games were pre-tested with five healthy adults and with five people with TBI. Based on pre-tests, it was possible to identify more specific inclusion and exclusion criteria for the study. In addition, specific games were identified and excluded, (eg, those which may cause dizziness or headaches in participants due to dark colors, 3D tunnel effects, and so on) [51].

Study participants will have full understanding of the content and purposes of the study. They will be informed orally a total of 3 times before their informed consent is asked for (2 telephone calls, 1 face-to-face meeting). Written consent to participate in the study will be requested from participants after they have been informed of all aspects of the study. According to the Finnish Medical Research Act (488/1999) [52] the participants will be informed that in case they withdraw from the study for any reason, their information that has already been collected will not be removed from the research material. Participants will have no obligation to provide information they do not want to share. They can also withdraw from the study at any time without providing a reason. Refusal to participate in the study will not have any effects on their daily lives. Further, the psychological safety and emotional stability of the responders also will be taken into account. They will be free to stop answering questionnaires at any time, in case they feel uncomfortable or distressed. If a participant would like to have additional information, they will be informed about a contact person who can answer their questions by telephone, email or text messaging.

To shorten the first face-to-face meeting with the Research Assistant, participants will receive paper-format questionnaires by post. For follow-up interviews (after the 8-week intervention and 3-month period) participants will return the filled questionnaires in a pre-paid envelope or during their face-to-face meeting with the Research Assistant. The participants in the "as usual" group will return the filled-in questionnaires by post

only. The participants will not be charged for any costs associated with data collection. Participants will be informed that if the console breaks down, they are not liable for damages. In special cases, the Technical Assistant will visit the participant's home to repair devices if needed. If any technological problems arise during the intervention, the participants have the opportunity to contact the Technical Assistant by phone or email to receive practical or hands-on support.

The privacy rights of human subjects will be observed at all times. Respondents' answers will not be provided to organizations outside the study. Information obtained will be used only for its original purpose. Information derived from research data will be coded and recorded in such a way that subjects cannot be identified [53]. All collected data will be transferred, recorded and securely stored (Personal Data Act 523/1999, Archive Act 831/1994, Constitutional Act 731/1999) [54-56].

The participants in the group assigned entertainment gaming may choose games targeted for an audience of young adolescents or 16 years old or younger. All participants in intervention groups can change the game during the intervention period if they find the game to be unpleasant, boring or otherwise dissatisfying. Moreover, all participants will be adults (over 18 years) who are capable of playing the games if they are willing to do so. The games being used are commercially available; however, some are designed only for adult players. These action games may include explicit graphic content, emotionally challenging themes or rough language. We found studies related to possible outcomes of playing this type of action game: some studies concluded that action games with violent elements may cause negative outcomes with aggression [57,58]. Contrasting results have also been found: it has been concluded, for example, that action games do not lead to aggressive behavior in adult users [59]. Regarding the potential positive effects of the games, they are associated with higher visuospatial cognition or visual skills [59]. Non-violent games do not appear to generalize to visuospatial cognitive abilities even when they involve visual rotation tasks [60].

The study will be conducted in accordance with the Finnish Medical Research Act (488/1999, amended 295/2004) [54], and Amendments (295/2004); Personal Data Act (523/1999) [54]; the Act on the Openness of Government Activities (621/1999). The principles of research ethics will be followed [61,62]. Any regulations addressing the conduct of a vulnerable population will be taken into account. This population could not be reached in another data collection method (eg, a register study).

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Conflicts of Interest

None declared.

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Abbreviations

ANOVA: analysis of variance
BRIEF-A: Behavior Rating Inventory of Executive Function – Adult Version
GSC: General Self-efficacy Scale
PASAT: Paced Auditory Serial Addition Test
PHQ-9: Patient Health Questionnaire
PS3: Playstation 3
TBI: traumatic brain injury
TEKES: The Finnish Funding Agency for Technology and Innovation
TMT: Trail Making Test
WAIS-IV: Wechsler Adult Intelligence Scale, 4th Edition

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Original Paper

Assessing the Feasibility of a Web-Based Weight Loss Intervention for Low-Income Women of Reproductive Age: A Pilot Study

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Abstract

Background: Low-income women of reproductive age are at increased risk for obesity and resulting increases in the risk of maternal/fetal complications and mortality and morbidity. Very few weight-loss interventions, however, have been targeted to this high-risk group. Based on the high prevalence of social media use among young and low-income individuals and previous successes using group formats for weight-loss interventions, the use of social media as a platform for weight-loss intervention delivery may benefit low-income women of reproductive age.

Objective: Examine the feasibility of delivering group-based weight-loss interventions to low-income women of reproductive age using face-to-face meetings and Web-based modalities including social media.

Methods: Participants attended a family planning clinic in eastern North Carolina and received a 5-month, group- and Web-based, face-to-face weight-loss intervention. Measures were assessed at baseline and 20 weeks.

Results: Forty participants enrolled, including 29 (73%) African American women. The mean body mass index of enrollees was 39 kg/m^2. Among the 12 women who completed follow-up, mean weight change was -1.3 kg. Participation in the intervention was modest and retention at 5 months was 30%. Returnees suggested sending reminders to improve participation and adding activities to increase familiarity among participants.

Conclusions: Engagement with the intervention was limited and attrition was high. Additional formative work on the barriers and facilitators to participation may improve the intervention's feasibility with low-income women of reproductive age.

(JMIR Res Protoc 2016;5(1):e30) doi:10.2196/resprot.4865

KEYWORDS

Obesity; Nutrition; Physical Activity; Minority Health; Healthcare Disparities; Intervention Studies; Internet; women; weight loss; mhealth

Introduction

Low-income women of reproductive age (WRA) are at increased risk for obesity [1], which is in turn associated with increased risk of maternal and fetal complications [2] during pregnancy, and morbidity [3] and mortality [4] throughout life. Few weight-loss intervention studies are targeted to low-income women [5]. Findings from recently reviewed individual- and group-format weight-loss interventions are encouraging, particularly group-based interventions [5]. To our knowledge, there is a dearth of studies investigating Web-based weight-loss interventions including social media as a mechanism to encourage social support and healthy behavior changes among low-income WRA.

Web-based delivery of intervention components may reduce barriers to intervention participation experienced by low-income WRA (e.g., lack of time or support) [6]. For example, social media websites (e.g., Facebook) may be effective social support platforms for weight-loss interventions among this population. These websites are used by 84% of Internet users aged 18-29 years, are more likely to be used by lower income Internet users, and offer group communications tools and the capacity for establishing networks of online friends [7]. Given these features, social media websites are potentially effective delivery mechanisms for group-based peer support interventions [8].

Although there is a growing body of research examining Web-based weight-loss interventions, [9] few have targeted low-income WRA and, to our knowledge, none have used social media with this group. The purpose of the current study was to examine the feasibility of delivering a previously tested group-based weight-loss intervention [10] adapted to low-income WRA using Web-based educational content and social media.

Methods

Study Design

This feasibility study employed a single group pre-post design. We piloted a weight-loss intervention among female participants enrolled from a county health department family planning clinic and assessed outcome measures at baseline and 5 months.

Recruitment

The Integrated Screening and Health Assessment, Prevention, and Evaluation (InShape) Study [11] screened 462 participants ages 18-44 years for cardiovascular disease risk factors. InShape eligibility criteria were being nonpregnant, English speaking, and attending an initial or annual family planning clinic visit at a health department located in eastern North Carolina. Each participant was given a brochure with information about enrolling in a weight-loss study or lifestyle study. Women who had a BMI greater than 27.5 kg/m^2 were considered eligible for the weight-loss study. Although the United States Preventive Services Task Force recommends that patients with a BMI greater than 30 take part in weight-loss interventions [12], those in the higher half of the overweight category (27.5 to 29.9) often wish to lose weight for both health and aesthetic reasons, and thus were invited to be in this study.

Participants who expressed interest in the weight-loss study were contacted by phone and invited to an enrollment visit. To have sufficient numbers for the group format component of the intervention, the intervention did not begin until all participants were enrolled. The Institutional Review Board of the University of North Carolina at Chapel Hill approved and monitored this study.

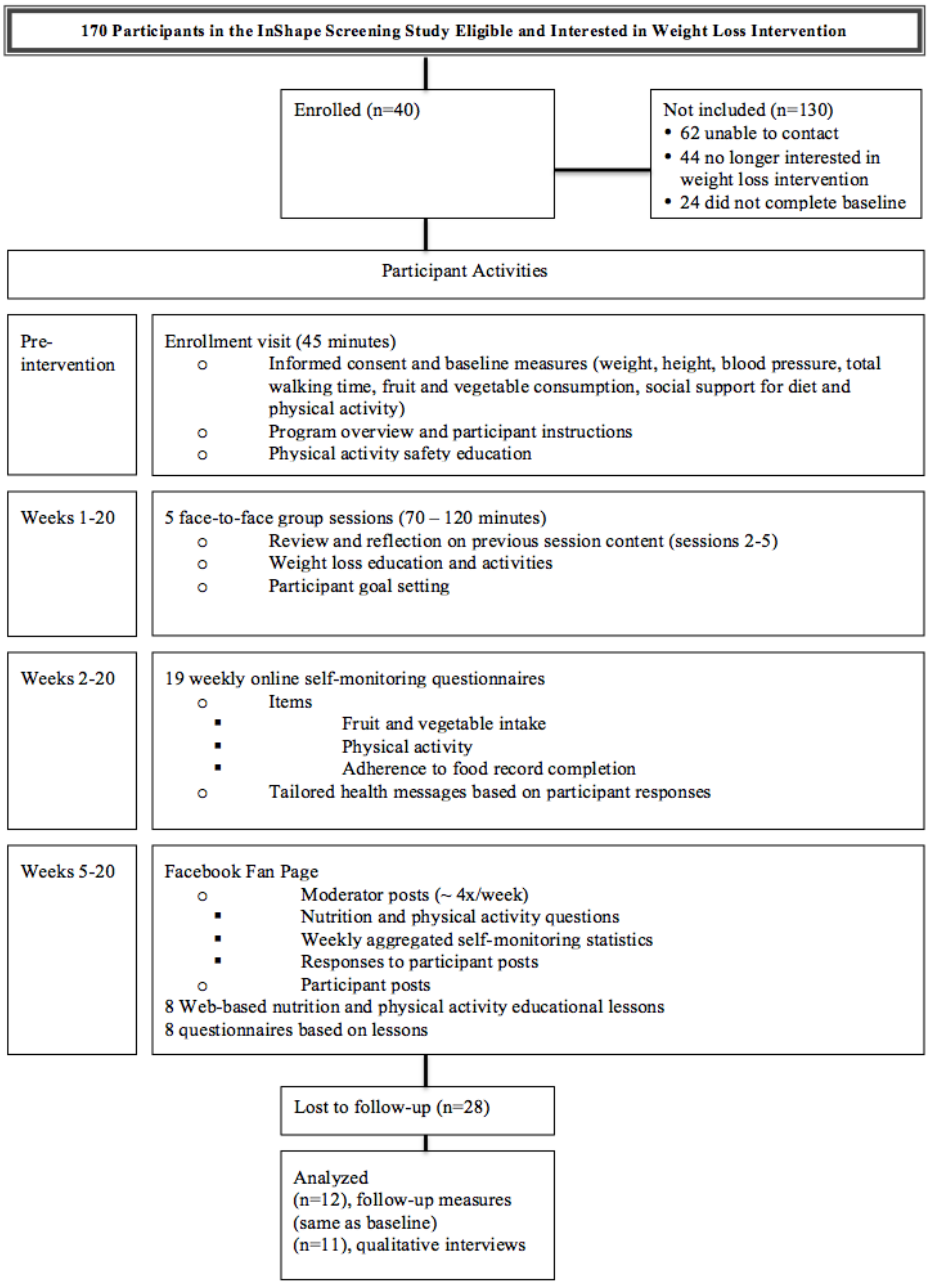
Program Adaptation

The InShape weight-loss intervention content was based on the Weight-Wise program, a group-based, 16-session, behavioral weight-loss intervention targeting low-income, midlife (40-64 years old) women [10]. The Weight-Wise program emphasizes goal setting, self-monitoring, feedback, and education to promote dietary and physical activity (PA) recommendations adapted from the Diabetes Prevention Program [13]. For this study, the Weight-Wise program was modified to be more feasible by maintaining the key behavioral components, decreasing the number of face-to-face sessions from 16 to 5, adapting content originally covered in the group sessions for delivery through Web-based modules, and including a social media component for social support.

Enrollment Visit

At the enrollment visit, conducted at the study research office, the interventionist first obtained informed consent and completed baseline measures (Figure 1). Then, during a 30 minute face-to-face, enrollment and counseling visit, the interventionist provided a program overview, instructed participants on how to perform the research study self-monitoring, provided PA safety information, and asked participants to set behavioral goals. A body weight scale, calorie counting book, and pedometer were provided to participants to take home.

Figure 1. Study flow diagram of participant activities.



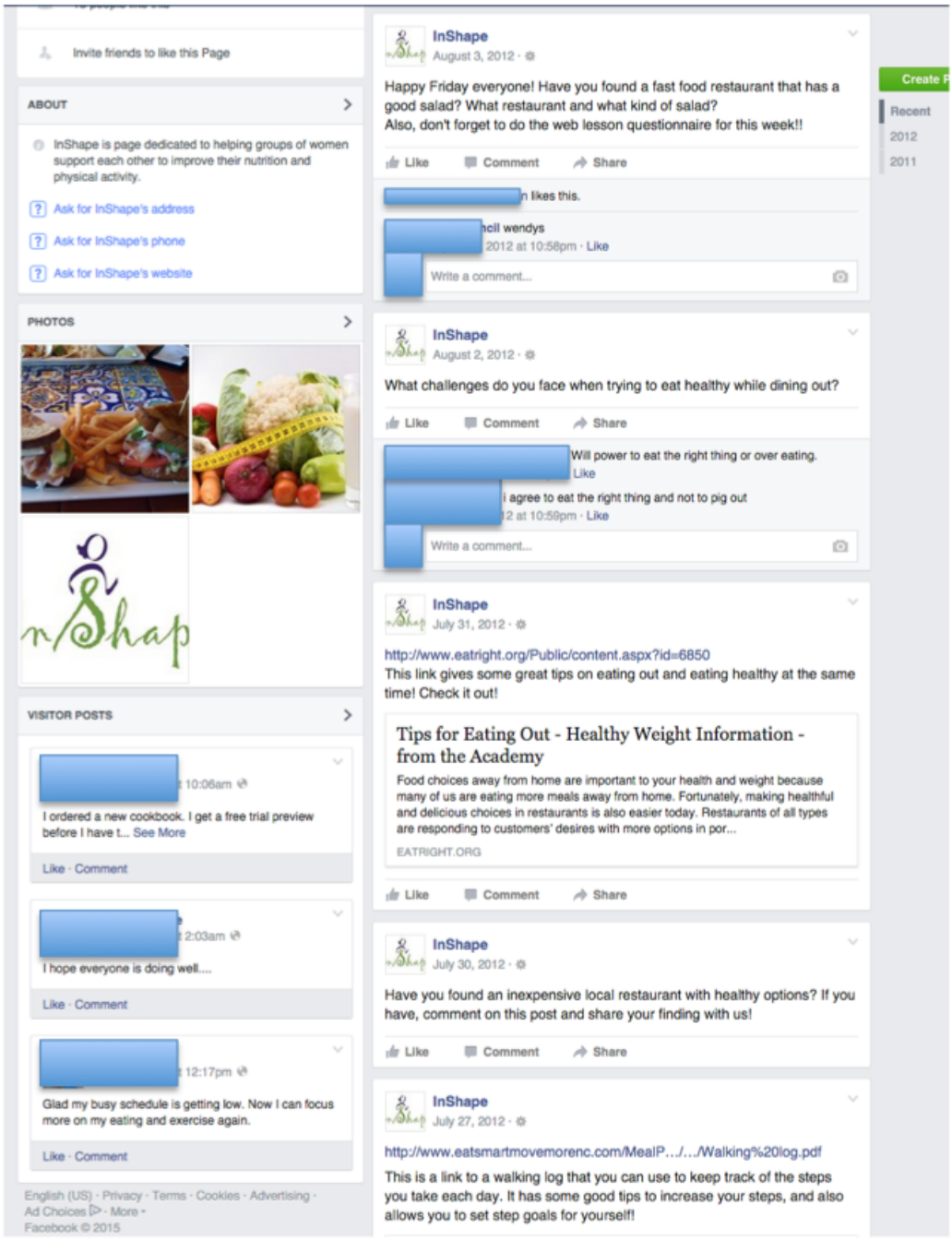
Group Sessions

Once a sufficient number of participants completed the enrollment and counseling visit, the remaining intervention activities were initiated. Five face-to-face group sessions (70-120 minutes) were held over a 20-week period at the health department on days and times that participants indicated were convenient (each session was offered 2 times during the week). The mean interval between the enrollment visit and the first group session was 85 (6-148) days. During each session, participants reflected on previous session content; received new intervention content, including hands-on activities such as PA or food preparation demonstrations; and completed an action plan for the coming week's goals.

Facebook Fan Page

At the onset of intervention activities, participants were invited to "like" a Facebook fan page created for the study, where participants could model healthy behaviors and support others in their dietary and PA behavior changes (Figure 2). Study staff "seeded" the fan page with weekly remarks designed to encourage participation, including ice breaker questions, questions related to lesson and group session content, and feedback about aggregate data from participants' weekly self-monitoring.

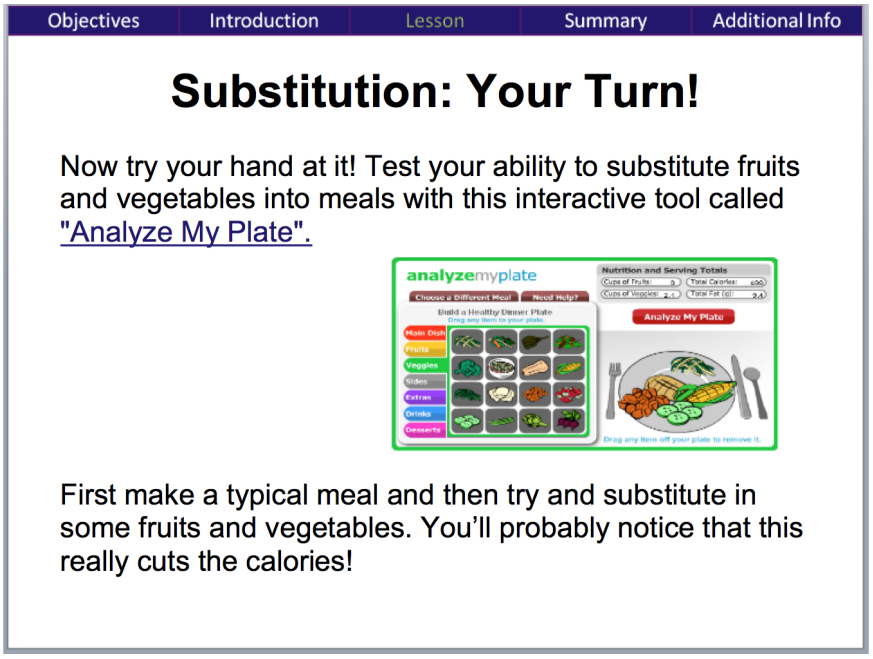
Figure 2. InShape Facebook Fan Page.



Web-Based Educational Lessons

Using Google docs, a Web-based document sharing program, we provided participants with hyperlinks (via email) to 8 Web-based nutrition and PA educational lessons every 2 weeks for 16 weeks, beginning 1 month after intervention activities were initiated (Figure 3). Each lesson was followed by an email invitation to complete a Web-based lesson questionnaire. These questionnaires asked participants about the topics covered in the Web-based lessons and provided reinforcing educational messages following each question. Lessons were independent (i.e., participants were not required to complete earlier lessons to participate in subsequent lessons).

Figure 3. InShape Web-based educational lesson.



Self-Monitoring with Tailored Feedback

The interventionist sent participants weekly emails containing a link to a brief, 6-question online self-monitoring questionnaire where participants were asked to self-report weight, frequency of keeping a food record, fruit and vegetable intake, amount of moderate physical activity, and number of steps. Tailored tips for improving or maintaining healthy behaviors were delivered to participants based on their questionnaire responses.

Qualitative Interviews

After completion of the intervention, we conducted 15-minute in-person and telephone qualitative interviews to assess intervention acceptability and barriers to participation. Incentives ($20 gift cards) were given at measurement visits (baseline and 5 months) and upon completion of qualitative interviews, and also were available based on "InShape points," which were earned by participating in intervention activities. We conducted 5 monthly drawings for points-based incentives. The number of entries/participant equaled the number of their InShape accumulated points.

Measures

Trained personnel collected data at baseline and 5 months. Weight and height (baseline visit only) were measured twice without shoes using an electronic scale and portable stadiometer and then averaged. Blood pressure was measured 3 times (after the participant was seated for 5 minutes and then at 1 minute intervals) with an Omron HEM-907XL automated blood pressure monitor and averaged.

We assessed multiple other outcomes through self-report at baseline and 5 months. Total walking time was documented with the RESIDE PA survey [14] and daily fruit and vegetable servings with a Block Rapid Food Screener [15]. Social support for diet and PA were measured using the Social Support and Eating Habits/Exercise scales [16]. Self-efficacy for diet and PA behavior change were each measured with a single question (eg, "On a scale of 1 to 5, how sure are you that you can walk or do a similar activity for 30 minutes or more on 5 or more days per week?").

Using the Qualtrics survey program (Qualtrics, Provo, UT) we recorded the frequency of participants' use of the online self-monitoring questionnaires and completion of the Web-based lesson questionnaires. The interventionist manually recorded Facebook fan page contributions and logged participant attendance at group sessions. At the end of the intervention period, participants also completed an acceptability questionnaire and a qualitative interview to solicit their thoughts about and experiences with the intervention and suggestions for improving it.

Statistical Analysis

Descriptive statistics were used to describe baseline characteristics and the frequency of participant involvement in intervention activities. Due to the limited sample size at 5 months, we did not use statistical tests to examine differences in measures at baseline and 5 months. Statistical analysis was performed with SAS software (Version 9.3, Cary, NC). Analysis of qualitative interviews included identifying key themes and quantifying instances of specific responses.

Results

Recruitment

Of the 251 (54%) participants in the InShape Screening Study who were eligible for the weight-loss intervention based on a BMI greater than 27.5 kg/m^2, 51 (20%) expressed an interest in the weight-loss study and 119 (47%) in either the lifestyle or the weight-loss study. Of these 170 eligible and initially interested participants, we successfully contacted 108 (64%). Of those contacted, 64 (59%) expressed continued interest in the weight-loss study with 40 (63%) completing the baseline visit to comprise the study sample.

Participant Characteristics

The majority of participants (N=40) were under 30 years of age, African American, low-income, and uninsured (Table 1). One-quarter of participants reported high blood pressure or had a measured systolic higher than 140 mmHg or diastolic greater than 90 mmHg and the average BMI was 39 kg/m^2. Over half were categorized as having class II or III (extreme) obesity. Most participants reported using the Internet (95%) and Facebook (83%).

Table 1. Baseline characteristics of female participants age 18-44 years (N=40).

Characteristic		n (%) or mean (SD)
Age, y, mean (SD)		30 (6.5)
Race [a]		
	African American	29 (73)
	White	10 (25)
Ethnicity [b]		
	Hispanic	5 (13)
Total annual household income		
	< $20,000	34 (85)
	$20,000-$39,999	5 (13)
	$40,000-$69,999	1 (3)
Currently employed full time		16 (40)
Health insurance		
	Private	2 (5)
	Medicaid	7 (18)
	No insurance	31 (78)
Education		
	Some high school	7 (18)
	High school graduate	19 (48)
	At least some college	14 (35)
Weight, kg, mean (SD)		106.0 (25.8)
BMI, kg/m^2, mean (SD)		39 (8.5)
	Overweight (BMI 25-29.9)	7 (17)
	Class I Obesity (BMI 30-34.9)	8 (20)
	Class II Obesity (BMI 35-39.9)	8 (20)
	Class III Obesity (BMI 40+)	17 (42)
High blood pressure (patient self-report, systolic ≥ 140 mmHg, or diastolic ≥ 90 mmHg)		10 (25)
High blood cholesterol (self-report)		4 (10)
Current smoker (self-report)		12 (30)
Diabetes (patient self-report of diagnosis or A1cValue ≥ 6.5)		4 (10)
Fruit and vegetables, servings/day, mean (SD)		3.3 (1.7)
Minutes walked, weekly, mean (SD)		59.1 (174.5)
Technology use		
	Use Internet	38 (95)
	Use Facebook	33 (83)

[a]Data for 1 participant were missing.

[b]Two participants did not report ethnicity.

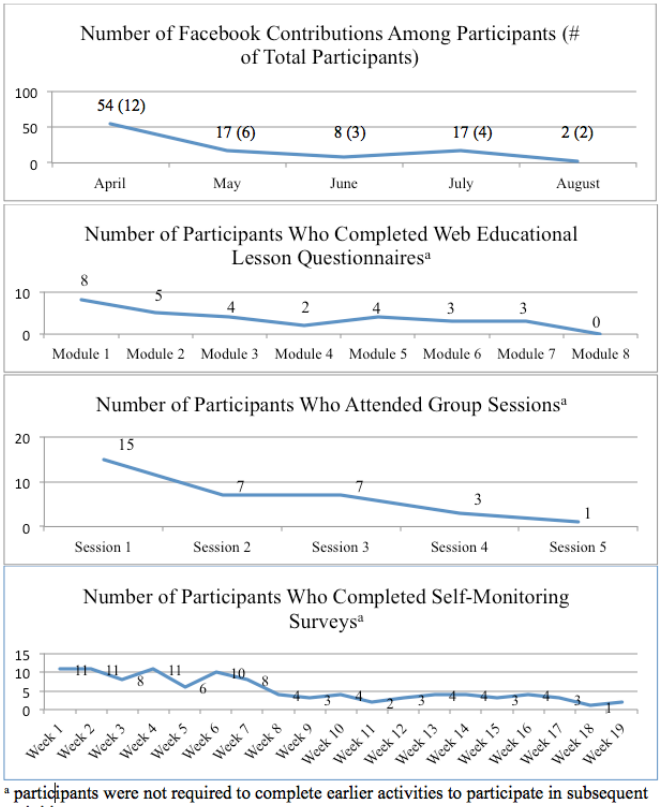
Participant Outcomes

Twelve participants (30%) completed 5-month measures (study returnees). At the end of the intervention period (5 months), the difference in mean weight among the 12 study returnees was -1.3 kg (SD=4.4 kg) with a range in weight change of -9 to 5 kg. Of note, 7 of 12 participants lost weight. Mean change in walking time was 116.3 minutes/week (SD=191.6 minutes/week) with a range of 0 to 675 minutes/week. Mean change in servings/day of fruit and vegetables was 0.5 servings/day (SD=1.5 servings/day) with a range of -2 to 3 servings/day.

Participant Use

Intervention activity participation was modest at the beginning and declined over time (Figure 4). A total of 12 participants contributed 98 comments to the Facebook group (mean=8, range = 1-20). Participants exchanged a variety of social support messages (eg, "Hi ladies I am looking for a walking partner I live in [City]" [response] "I live too far for an everyday thing...but we could always meet at a park! Lemme know!") Completion of Web lesson questionnaires declined from 8 women (module 1) to none (module 8). Eighteen women participated in 1 or more face-to face group sessions. Completion of self-monitoring questionnaires declined from 11 (week 1) to 1 (week 18).

Figure 4. Participation in intervention activities (N=40).



[a] participants were not required to complete earlier activities to participate in subsequent activities

Participant Evaluation of Intervention

Qualitative analysis (n=11) suggested that participants had hectic lives and work schedules that interfered with intervention participation and behavior change. Some returnees reported lack of access to the Internet, lack of transportation, and a desire for additional tools like digital calorie counting applications and food scales. Returnees wanted more frequent group sessions and more meeting times and suggested more text and email reminders. Suggestions for improving the Facebook component of the program included having team building exercises, enlisting friends, and providing a list of participants to each person to facilitate more face-to-face interaction with other participants. Participants also suggested more moderator posts, reminders, and greater frequency of providing incentives.

Discussion

Despite the need for weight loss among our sample of overweight and obese participants, only a small percentage were interested in joining the study and 30% of participants completed the intervention. On average, minimal changes in weight were observed among returnees.

Principal Findings

Use of Web-based components was low and eroded over time. Despite the high levels of self-reported Internet and social media use, it is possible that participants' Internet access was episodic rather than continuous, which may have hindered participation in Web-based components. Lack of familiarity with other participants prior to using the Facebook fan page may have limited engagement in the social media intervention component. Attrition in the current study was greater than previous

Web-based weight-loss intervention studies that mostly included older populations, including some that used Facebook for intervention delivery [17,18]. Participation was low for group sessions as well, indicating that there are likely reasons unrelated to mode of delivery that affected engaging in the intervention. The long interval between expressing interest and beginning study activities (mean 85 days) may have exacerbated traditional barriers to participation that arise over time (eg, moving frequently). This delay was based on our desire to have a "critical mass" of participants for the social media component at the outset and to have the components delivered in concert.

Future social media interventions should plan for this contingency by having rapid recruitment procedures in place. It may also be necessary to provide some intervention components or other content to participants in this lag period to maintain interest. In addition, having multiple intervention components on different platforms (ie, Facebook, Google, email, and Qualtrics) may have produced too much burden for participants. Retention could possibly be improved by delivering all intervention components through 1 platform. Other factors endemic to our study population may have also played a role in limited engagement and high attrition. For instance, young adults cite different weight loss motivations (eg, appearance and social pressure) compared to older adults (eg, health concerns) [19]. Our emphasis on the health benefits of weight loss may not have resonated with young women and, given nearly 75% of the sample was African American, greater normative acceptance of higher BMI may have limited the effectiveness of our intervention [20].

Several suggestions for improving levels of engagement in future studies targeting low-income WRA can be drawn from our results. Based on participant feedback about the need for reminders and the high penetration of cellular phones in low-income populations, [21] incorporating text message reminders or content could improve participation. Our qualitative data also suggest that social media-based interventions should build familiarity between participants prior to intervention activities. This could be accomplished by establishing stronger ties between intervention participants or incorporating existing online social ties (eg, friends, family) into social media interventions. Screening for participant motivation, which was not done in this study, has been undertaken in previous weight-loss intervention studies that reported greater participation and retention [10,22]. Finally, weight-loss intervention content should be tailored to factors that are motivating for young adults, such as physical appearance and social factors [19].

Limitations

This study has several limitations. There was a significant lag between many participants expressing interest and beginning the intervention. Given the small number of returnees, we did not undertake statistical testing. Misclassification is possible since we measured PA by self-report, which is subject to overreporting, [23] and our measure of Internet access did not differentiate between episodic and continuous access. Nonreturnees, who may have had less positive attitudes toward the intervention or expressed additional barriers to participation, were not included in qualitative interviews. We recommend that future intervention research targeting populations with traditionally high attrition develop a priori strategies for collecting data from nonreturnees who may be in the best position to explain the most salient factors associated with their lack of participation.

Conclusions

Although use of the Web and social media in particular is prevalent among young, lower income women, participation in the Web-based components of our intervention was limited. Additional formative research with larger samples of low-income WRA should be conducted in order to develop novel intervention strategies for weight loss among low-income women of reproductive age who are at high risk for both chronic disease and poor pregnancy outcomes.

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Conflicts of Interest

None declared.

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Abbreviations

InShape: Integrated Screening and Health Assessment, Prevention, and Evaluation
PA: physical activity
WRA: women of reproductive age

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Original Paper

A Community-Engaged Approach to Developing a Mobile Cancer Prevention App: The mCPA Study Protocol

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Abstract

Background: Rapid growth of mobile technologies has resulted in a proliferation of lifestyle-oriented mobile phone apps. However, most do not have a theoretical framework and few have been developed using a community-based participatory research approach. A community academic team will develop a theory-based, culturally tailored, mobile-enabled, Web-based app—the Mobile Cancer Prevention App (mCPA)—to promote adherence to dietary and physical activity guidelines.

Objective: The aim of this study is to develop mCPA content with input from breast cancer survivors.

Methods: Members of SISTAAH (Survivors Involving Supporters to Take Action in Advancing Health) Talk (N=12), treated for Stages I-IIIc breast cancer for less than 1 year, 75 years of age or younger, and English-speaking and writing, will be recruited to participate in the study. To develop the app content, breast cancer survivors will engage with researchers in videotaped and audiotaped sessions, including (1) didactic instructions with goals for, benefits of, and strategies to enhance dietary intake and physical activity, (2) guided discussions for setting individualized goals, monitoring progress, and providing or receiving feedback, (3) experiential nutrition education through cooking demonstrations, and (4) interactive physical activity focused on walking, yoga, and strength training. Qualitative (focus group discussions and key informant interviews) and quantitative (sensory evaluation) methods will be used to evaluate the participatory process and outcomes.

Results: Investigators and participants anticipate development of an acceptable (frequency and duration of usage) feasible (structure, ease of use, features), and accessible mobile app available for intervention testing in early 2017.

Conclusions: Depending on the availability of research funding, mCPA testing, which will be initiated in Miami, will be extended to Chicago, Houston, Philadelphia, and Los Angeles.

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KEYWORDS

smartphone applications; African Americans; breast cancer survivors; cancer prevention guidelines; dietary intake; physical activity

Introduction

For women in the United States, breast cancer is the most common cancer and the second leading cause of cancer death [1]. Although white women have historically had higher incidence rates than African American (AA) women, the rates converged in 2012 [2]. Breast cancer death rates are higher for AA women compared to white women [3]. AA women are more likely to be diagnosed with breast cancer at younger ages and with more aggressive and advanced tumors. Although breast cancer survival in AA women has increased, survival rates remain lower than among white women [4].

Modifiable lifestyle risk factors related to energy balance may contribute to racial/ethnic disparities in breast cancer survival rates. Disparities in these factors are large and persistent, particularly between white and AA women. Recent data from the Behavioral Risk Factor Surveillance System revealed three disparity risk categories for AA women: (1) obesity (35.7% vs 23.7% for whites), (2) inadequate fruit and vegetable consumption (12.6% vs 17.4% for whites), and (3) low physical activity (63.8% vs 50.3% for whites) [5]. Lifestyle behavioral guidance to promote health and prevent disease is a cornerstone of American public health policy, yet relative to women of other races, AA breast cancer survivors are less likely to report adherence to the American Cancer Society [6] and the American Institute for Cancer Research (AICR) [7] cancer prevention guidelines.

Limited research on AAs exists because they have been underrepresented in studies examining health behaviors that improve breast cancer survival. One of the few studies with AA women, the Women's Healthy Eating and Living Study, showed that at baseline, AA survivors were more likely to be obese (45% vs 25% for whites), to consume more calories from fat (+3.2%), to have fewer servings of fruits (-0.7/day), and were less successful at making and maintaining dietary changes than whites [8]. Greenlee et al [9] completed a randomized controlled trial with the commercially available Curves program, following 42 Hispanic and AA breast cancer survivors (BCSs) for 6 months. The trial resulted in weight loss that was not maintained at 6 months after the intervention. A community-based pilot study of 24 AA BCSs engaged in walking as physical activity (PA) [10] resulted in increases in steps walked per day and decreases in body mass index (BMI), body weight, and waist/hip circumferences, with most changes maintained at 3 months. A pre-post design, that included one of two weekly sessions dedicated to exercise, was used to test a 6-month intervention with 23 AA BCSs [11]. Participants experienced changes in weight, BMI, and social support. In a 16-week home-based motivational exercise program for 13 AA BCSs, there was a post-intervention increase in total minutes of PA and improved physical functioning [12].

To enhance our understanding of health behaviors that improve survival among AA BCSs, a lifestyle needs assessment of 240 members of the support group, SISTAAH (Survivors Involving Supporters to Take Action in Advancing Health) Talk, was completed. Most BCSs reported lack of adherence with cancer prevention recommendations for portion control (89%), body weight (68%), PA (83%), and intake of vegetables/fruits (75%), processed meat (54%), and red meat (53%) [13].

SISTAAH Talk members expressed interest in developing an alternative format for a lifestyle intervention. Rapid expansion of mobile technologies, including smartphone apps, provides such an opportunity. Currently, 85% of American adults own a mobile phone, and 45% own a smartphone, with usage higher among AAs (47%) relative to whites (42%) [14]. Half of all smartphone owners use their phones to search for health information, with 60% of all downloaded health-related apps involving weight loss and exercise [15].

A variety of apps relating to diet, nutrition, and weight control are available from platforms such as iPhone, Android, Nokia, and BlackBerry [16]. Traditional lifestyle interventions are facilitated and resource-intensive and may or may not be evidence-based. mHealth apps are mediated, less expensive, and rarely grounded in theory or evaluated using scientific methods [17-19]. To our knowledge, however, there are no apps that promote adherence to science-based cancer prevention guidelines. Adherence to these guidelines to prevent breast cancer recurrence is the primary focus of the Mobile Cancer Prevention App (mCPA) and such compliance may, in turn, contribute to improving health-related quality of life (eg, physical functioning, depression) [20]. Investigators postulate that employing principles of community-based participatory research (eg, unit of identity, building on resources within the community, cyclical and iterative processes) in developing a smartphone app, will result in widespread use and broader acceptance of cancer prevention guidelines. The purpose of this study is to partner with members of a breast cancer support group to develop a cancer prevention app for subsequent testing in a behavioral intervention study. Investigators hypothesize that a participatory process, one that engages individuals affected *by* the problem in developing solutions *to* the problem [21], will result in an acceptable (frequency and duration of usage), feasible (structure, ease of use, features), and accessible mobile cancer prevention app.

Methods

Study Population

Established in 1995 as the first breast cancer support group for women of color in South Florida, the goal of SISTAAH Talk is to provide a forum for African-Americans to communicate about and make sense of their diagnosis and treatment in order to achieve improved physical and mental health outcomes. SISTAAH Talk includes women from across Miami-Dade and Broward counties, reaching an average of 20 women each month through education, outreach, and research. In 2013, SISTAAH Talk breast cancer survivors (N=240) participated in focus group discussions and completed lifestyle assessments for the National Institute of Minority Health & Health Disparities-funded pilot study, Assessing Lifestyle Modification Needs & Experiences of African American Women [13]. Four focus group discussions (n=42; mean age 45.73 years, SD 7.91, range 35-75 years) identified barriers to and intervention approaches for enhancing dietary intake and PA. Themes emerging from content analysis converged into the following categories: practical tools (goal

setting, self-monitoring), talk as central, led by BCSs, support group approach, "hands-on" or interactive nutrition (cooking) and physical activity education, and community-based (not community placed) research. The findings of these focus group discussions will be combined with theories in an intervention planning process between investigators and BCSs to inform the design of mCPA.

SISTAAH Talk members (n=12), treated for >1 year for Stages I-IIIc breast cancer, 75 years of age or younger, and English speaking/writing, were identified by leaders of the support group as good role models to participate in developing mCPA. Each applicant was interviewed by the principal investigator and support group facilitator to determine comfort level in participating in focus group discussions, key informant interviews, and app content development (eg, videotaping cooking demonstrations and PA).

Participatory Engagement

SISTAAH Talk members participating in mCPA reviewed themes from focus group discussions during three telephone support group meetings with the principal investigator. To describe the participatory process of app development, Table 1 outlines contributions for each element used in the process.

Table 1. Participatory process of app development.

Element	Participant contribution	Investigator contribution	Outcome
Theoretical framework	Consider importance of lifestyle change to everyday life and survival	Identified theory to undergird BCSs' belief that eating healthy and exercising regularly impacts survival	Health Belief Model; Theory of Planned Behavior
Peer-led activities	Include SISTAAH Talk members in all activities	BCSs as equal partners (facilitators, discussants) of cooking demonstrations, PA sessions, and focus group discussions	12 BCSs selected by SISTAAH Talk facilitator and mCPA principal investigator
Hands-on experiences	Mirror SISTAAH Talk activities by featuring BCSs cooking and exercising	Experiential nutrition and PA education	YouTube videos
Dietary intake	Feature recipes developed by AA community members; include practical, achievable recommendations that avoid drastic changes	Strategies and recipes promoting Cancer Prevention Guidelines	Down Home Healthy Living Cookbook
Physical activity	Walking, yoga, and strength training; advice from a BCS; consider different body sizes and practical, achievable goals	Symptoms such as lymphedema (swelling in an arm), arthralgia (pain in a joint), and neuropathy (numbness or weakness)	BCS-led walking, yoga, and strength training PA
Talk as central	The SISTAAH Talk approach to discussing lifestyle change	Incorporate beliefs, benefits, and barriers to lifestyle change as an instructional tool	Didactic sessions for goals, monitoring progress, and providing/receiving feedback

Theoretical Framework

To provide a conceptual basis to undergird mCPA content, we will use the (1) health belief model (HBM) and (2) theory of planned behavior (TPB) (see Table 2). The HBM suggests that health-related cognitions for determining behavior considers the breast cancer survivor's *belief* that lifestyle behaviors affect breast cancer recurrence, how *severe* the recurrence would be, and the *cost/benefits* of lifestyle change [22]. The TPB posits that health behavior is affected by past breast cancer experience and social norms (ie, lifestyle practices) more so than beliefs (ie, a link between breast cancer recurrence and lifestyle) [23].

Table 2. Theoretical framework by mCPA construct.

	Health Belief Model[a]			Theory of Planned Behavior[b]	
Component	HBM1	HBM2	HBM3	TPB4	TPB5
Education	×	×	×		
Instructions			×		
Goal setting					×
Social support				×	
Provide feedback					×
Prompt review					×
Self-monitoring		×			×
Teach use of cues		×			
Action planning				×	

[a]Health Belief Model: HBM1 (perceived costs); HBM2 (health benefits); HBM3 (cues for action).

[b]Theory of Planned Behavior: TPB4 (subjective norms/social support); TPB5 (behavior control).

Cancer Prevention Guidelines and App Content

The proposed app will focus on the AICR guidelines for cancer survivors [7]: (1) Be as lean as possible without becoming underweight, (2) Be physically active for at least 30 minutes every day to help prevent cancer and prevent recurrence of cancer, (3) Avoid sugary drinks and limit consumption of energy-dense foods (particularly processed foods high in added sugar, low in fiber, or high in fat), (4) Eat more of a variety of vegetables, fruits, whole grains, and legumes such as beans, taking up at least 2/3 of your plate, (5) Limit consumption of red meats (such as beef, pork, and lamb) and avoid processed meats, taking up only 1/3 or less of the space on your plate, (6) If consumed at all, limit alcoholic drinks to two for men and one for women a day, and (7) Limit consumption of salty foods and foods processed with salt (sodium) by substituting herbs and spices high in phytochemicals (eg, basil, turmeric, paprika, thyme, and dill).

mCPA will include a flexible and autonomous approach to changing eating and PA behaviors. Tools on the app to support the development of self-regulatory skills and successful lifestyle change strategies will feature a range of instruments (eg, informational and self-monitoring) designed to enhance users' awareness of and motivation to work toward cancer prevention guidelines. For example, informational tools will include options to view individual goals and plans and to access selected strategies (eg, eating out, planning meals, incorporating PA through the day). Self-monitoring tools will include options to receive personalized feedback on progress and access to support through links to social media (eg, Facebook, twitter). App users will establish dietary intake and PA goals, access strategies to support these goals, develop plans of how to meet them, review progress, and receive feedback on goal achievement. Employing a support group format, users will access cooking demonstrations, exercise instructions, and practical advice through links to YouTube videos.

A community-engaged process for transforming main dishes, side dishes, snacks, and desserts into healthier options and for presenting advice on dietary intake and physical activity for cancer prevention has been previously described [24]. Dishes from the cookbook with lifestyle tips, entitled, "Down Home Healthy Living (DHHL) 2.0," [25] will be featured on the app. The purpose of the cookbook is to promote awareness of cancer prevention guidelines in African American communities. For the DHHL 2.0 cookbook with lifestyle tips, recipes were solicited from the National Black Leadership Initiative on Cancer community coalitions and dietary intake advice from participants in the Educational Program to Increase Colorectal Cancer Screening (EPICS). With guidance from a chef and registered dietitian, recipes were tested, assessed, and transformed. Lifestyle advice was obtained from focus groups. The cookbook has been distributed in print form to 2500 EPICS participants and shared electronically through 25 websites.

Physical activities for the app, specifically, walking, yoga, and strength training, were selected by SISTAAH Talk members during focus group discussions as options for inclusion in an intervention. A SISTAAH Talk member, BCS, and certified, licensed, insured fitness instructor will lead the PA experiential and instructional videos.

Mobile Cancer Prevention App Development

A timeframe for developing the app is included in Table 3. The initial step in developing the app was establishment of a research protocol. SISTAAH Talk members met with investigators via telephone conference call August-September 2015 to complete the research protocol for obtaining Institution Review Board (IRB) approval, which was granted in October 2015. The Georgia Regents University IRB approved this research plan. Informed consent will be obtained from all participants.

During August-September 2015, content and format for the app was also outlined. SISTAAH Talk members will be videotaped during cooking and PA demonstrations at three time points (December 2015, January and February 2016). A prototype of the app will be developed, presented, revised, and tested during the final 6 months of the study (May-October 2016).

Table 3. App development timeline.

Steps	Oct. 2015	Dec. 2015	Jan. 2016	Feb. 2016	Mar. 2016	Apr. 2016	May 2016	June 2016	July 2016	Aug. 2016	Sept. 2016	Oct. 2016
Obtain IRB approval	✓											
Outline app content	✓											
Conduct focus groups		✓	✓	✓								
Tape cooking demos		✓	✓	✓								
Tape PA demos		✓	✓	✓								
Analyze focus group data					✓	✓	✓					
Conduct key informant interviews		✓	✓	✓								
Analyze key informant interview data					✓	✓	✓					
Develop app prototype								✓	✓			
Pretest draft app										✓		
Finalize app											✓	
Publish manuscripts				✓				✓				✓
Submit research proposals												✓

The proposed Web-based app will run like a Web page, with the app operating on an external server and the user accessing the app through the Web browser from a smartphone. The app will be developed and prototype-tested prior to going live on the smartphone. Participants will be provided a link to the app, which will transition from gray to color, with a pop-up message indicating that a component is now available for feasibility testing. Figure 1 provides a sample of the smartphone app interface.

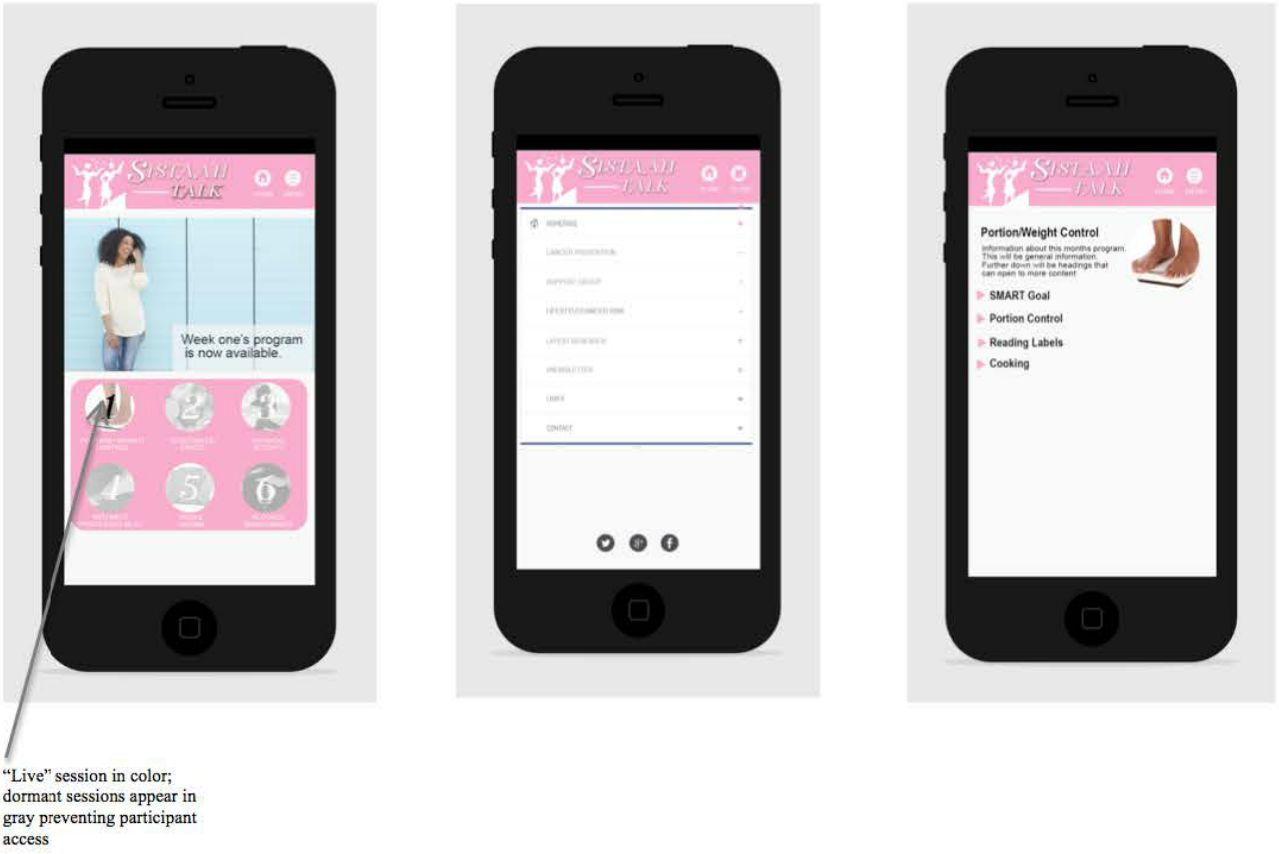
Following informed consent, participants will engage in four activities to develop the content of the app. Each activity will be audio- and videotaped, with edited versions serving as content for mCPA. The first activity, didactic instructions, will provide information on goals for, benefits of, and strategies for enhancing dietary intake and PA. Researchers will present evidence linking lifestyle to breast cancer incidence and recurrence, review AICR guidelines for cancer survivors, and provide strategies for adhering to these guidelines, and address questions and concerns of the participants. Next, through interaction with investigators and other support group members, participants will develop app features for setting individual SMART (specific, measurable, actionable, realistic, time-sensitive) goals, monitoring progress, and providing/receiving feedback. Experiential nutrition education, with hands-on activities, including recipe development, modification, and taste testing through interactive cooking demonstrations, will be captured for the app. Finally, participants will engage in interactive PA, including walking at various levels (eg, power, speed interval, walking-to-jogging, and stretching), yoga (eg, physical postures, conscious breathing, and meditation), and strength training (eg, slowly progressive lifting of lightweight dumbbells) though a series of 30-minute sessions.

Focus group discussions and key informant interviews will be conducted to obtain participants' feedback about app usefulness, identify the need for new system features and design requirements, and measure the acceptance of the mobile app and its features. Questions have been developed for the focus group discussions and key informant interviews based on the health belief model and theory of planned behavior (see Multimedia Appendix 1).

Four 90-minute focus groups with 12 SISTAAH Talk members and led by investigators, will explore three factors: performance expectancy, effort expectancy, and participant-centered factors. Performance expectance is defined as the degree to which a BCS believes that using the app will help attain lifestyle goals. Effort expectancy is the degree of ease associated with use of the app. Participant-centered factors include developing an app that is respectful of and responsive to user preferences, needs, and values, and ensuring that BCSs guide the development process. Acceptability will be determined by attitudes towards technology, anxiety, self-efficacy, and behavioral intention (eg, intent to use the app). Usability will be measured based on facilitating conditions and user-friendliness.

Three key informant interviews for each participant (n=36) will be conducted to ensure that perspectives across mCPA users are captured. Participants will be charged at baseline, midway, and postdevelopment with reviewing the app to make suggestions for adaptations and refinements and determine acceptability. Key informants will be provided access to the app, as it is being developed, and asked for feedback as part of a telephone follow-up.

Figure 1. A sample of the mobile app interface before usability testing.



Data Collection

Tasting samples or prepared dishes specific to the cancer prevention guidelines (eg, replace refined grains with whole grains, reduce red and processed meat consumption, consume at least 5 fruits and vegetables per day, reduce portion sizes to lower body weight, and lose even small amounts of body weight) will be distributed. Participants will complete a sensory evaluation of the appearance, taste, texture, aroma, and overall acceptability of dishes prepared during cooking demonstrations. With a Likert scale, participants will be asked to rate each dish from 1 (unattractive; flavor did not appeal to me; inappropriate texture; unappetizing aroma; unacceptable) to 5 (extremely attractive; tasted great; great texture; smelled good; extremely acceptable). A discussion of the sensory evaluation results at the end of the cooking videos will be used to show app users the acceptability of the featured dishes.

Focus group discussions and responses to key informant interviews will be digitally recorded, transcribed verbatim, manually coded, and summarized. Data will be analyzed using Qualitative Content Analysis [26]. Coding steps will include developing preliminary themes creating additional codes based on themes that arise, developing non-substantive codes, and producing detailed codes for analysis of specific topics. NVivo 10 software for computer-assisted qualitative data analysis will be used to facilitate the coding process (ie, assessing the degree of agreement/disagreement across themes and calculating interrater reliability scores) [27]. A process of double coding (eg, 2 coders will code all items) will be utilized to calculate a level of agreement between coders and to determine consistency in coders. Recurring themes will be identified, the research team will come to a consensus on coded themes, and themes will be summarized for analysis.

After themes are applied, the first iteration of mCPA will be presented in a discussion forum. The discussion forum will include all SISTAAH Talk members attending monthly meetings, which will include the mCPA participants. All data related to developing the app and the app itself will be presented to determine acceptability (intention to use). Based on comments of the participants during the discussion, a second iteration of the app will be completed and made ready for testing. The second iteration of the app will be reviewed by the technology expert, principal investigator, and support group leaders, who will decide that the app is ready for pilot testing. This process will strengthen content validity and permit user input into intervention development.

Data Analyses

mCPA acceptability, feasibility, and accessibility will be evaluated using descriptive statistics of frequency and proportion for discrete data and means and standard deviations for continuous data. All analyses will be accomplished with SAS version 9.4.

Results

It is anticipated that development of an acceptable (frequency and duration of usage), feasible (structure, ease of use, features), and accessible mobile app will be available for intervention testing in early 2017.

Discussion

Principal Considerations

We expect the mobile-enabled, Web-based app (mCPA), which is theory-based and culturally tailored, to lead to multicenter, randomized controlled trials of its effectiveness in promoting healthy dietary intake and physical activity among African-American breast cancer survivors. An effective, research-tested mobile intervention to assist survivors in adopting and maintaining healthy behaviors would fill a gap in the current evidence based on culturally appropriate, low-cost interventions that can help African-American women maximize their health and quality of life following a breast cancer diagnosis. The availability of effective mHealth interventions for survivors would allow for future dissemination and implementation in order to reach large numbers of women at a relatively low cost.

Apps for patient self-management, such as the mCPA, empower women to take control of their own health and come to terms with what is often a frightening and anxiety-provoking diagnosis. The use of new technologies, apps, and social media for patient self-management of their illness and to reduce the risk of recurrence can be contrasted with computer-based programs that allow for providers to communicate with their patients as part of breast cancer treatment and survivorship care. The latter are closely tied to clinical care and may not be accessible or affordable to all groups of breast cancer survivors.

Conclusions

Depending on the availability of research funding, mCPA testing, which will be initiated in Miami, will be extended to Chicago, Houston, Philadelphia, and Los Angeles. Results from the current study may lead to refinements, such as developing culturally tailored components of the app for ethnic subgroups of US blacks (eg, English-speaking Haitian-, and Caribbean-born blacks).

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Conflicts of Interest

None declared.

Multimedia Appendix 1

Topic and interview guides.

[PDF File (Adobe PDF File), 29KB - resprot_v5i1e34_app1.pdf]

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Abbreviations

AA: African-American
AICR: American Institute on Cancer Research
BCSs: breast cancer survivors
BMI: body mass index
DHHL: Down Home Healthy Living
EPICS: Educational Program to Increase Colorectal Cancer Screening
HBM: Health Belief Model
IRB: Institutional Review Board
mCPA: mobile Cancer Prevention App
PA: physical activity
SISTAAH: Survivors Involving Supporters to Take Action in Advancing Health
SMART: specific, measurable, actionable, realistic, time-sensitive
TPB: Theory of Planned Behavior

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Viewpoint

Patient Recruitment 2.0: Become a Partner in the Patient Journey Using Digital Media

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Abstract

We describe a digital platform, Pioneering Healthcare, designed to inform and empower people who are impacted by lung cancer. The platform enables Roche to support an online conversation with patients and caregivers about lung cancer, and about the role of lung cancer clinical studies in the development of future treatment options. This conversation is live and ongoing on the platform. It provides insights about the views and motivations of patients, and about how to better support patients pursuing treatment for life-threatening illness. We discuss the strategies used to deploy Pioneering Healthcare, and the advantages of using digital platforms for raising disease awareness, increasing patient engagement and, ultimately, for boosting patient enrollment into clinical trials.

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KEYWORDS

Internet intervention; patient empowerment; cancer survivor; systematic review; web 2.0; social media

Introduction

Establishing a digital platform that can deliver a benefit for patients and their caregivers requires insights about the severe psychological impact of receiving a diagnosis of late stage lung cancer. The impact of such a diagnosis extends beyond the patient and includes family members and caregivers. The messages and general presentation of any platform intended to support this group of individuals must acknowledge the significant sense of anxiety, grief, and loss that is experienced by individuals in such situations [1,2]. To deliver a genuine and unique benefit to visitors of our platform we must first understand these experiences, and follow best practices in the selection of informational content that we deliver to this vulnerable audience. Our ongoing research focuses on how we can support people emotionally, and with practical information, as they navigate their own personal way forward.

When detected at early stages lung cancer may be treated with curative intent using surgery, conventional chemotherapy, and radiotherapy. However, at later stages of the disease a complete remission may not be possible using standard treatment. Patients and caregivers may lose hope of finding a path toward recovery. Following a late stage lung cancer diagnosis, patients and family caregivers exhibit depression and anxiety at levels exceeding those observed in the general population [3,4]. Recent studies highlight significant benefits of psychotherapeutic intervention for this patient group as a means to ameliorate their distress, and perhaps even to improve therapeutic outcomes [5,6]. Beyond

the strictly clinical setting there are benefits, and unmet needs, associated with the provision of information and psychological support for patients and caregivers [1,7-9]. Organizations such as the American Cancer Society and Cancer Research UK provide this kind of support through patient advocacy and disease awareness programs.

Digital and online activities are becoming increasingly significant in the provision of support to patients. Indeed, people confronting a serious medical diagnosis have been going online in search of emotional support and medical information for as long as the Internet has existed. The Pioneering Healthcare platform seeks to prepare and support people to make decisions about their own unique way forward as they face advanced cancer. It seeks to achieve this using an online platform that could potentially reach any patient or caregiver with access to the Internet and bring them in contact with the experiences of other people impacted by lung cancer. Accordingly, we publish a combination of basic medical information as well as content that is designed to provide visitors with psychological support and practical information [10]. We author succinct descriptions of medical aspects of the disease including information about diagnosis and treatment. We also clearly define the purpose of and the processes involved in enrolling in a clinical trial. Beyond the succinct scientific content, the platform addresses psychological and practical goals by featuring practical content about finding support services and advice about communicating with family and friends. Importantly, we enable visitors to share their insights with other visitors to the website by publishing individual messages. We also enable visitors to post comments about messages that have been contributed by other visitors.

Through the platform we seek to raise awareness of late stage lung cancer and what it means to participate in a lung cancer clinical trial. As such, basic clinical trial processes are described including eligibility and exclusion. We describe how clinical trials provide access to investigational treatments, and how this access differs from the access that patients have to standard treatment options. Finally, we provide the telephone contact information of Roche's Trial Information Support Line based in North America. Callers can use this to reach trained health care professionals, who are able to answer questions about how to enroll in Roche-sponsored clinical trials. The site can be adapted rapidly to accommodate evolving changes to Roche's clinical development program, such as the initiation of a new clinical trial.

The platform links to additional digital resources including a mobile app and an instructional video for people recovering from thoracic surgery. These resources have been produced specifically for the platform and have been reviewed by internal and external health care professionals. Pioneering Healthcare also features a blog, a music channel on the online music streaming service Spotify, and online polls. The polls enable surveys to be conducted on topics relating to cancer treatment. Patients can contribute their views quickly and conveniently. They are then presented with the poll results to help them gauge the views of the broader online community. Various components of the platform are summarized in Figure 1.

In addition to keyword search advertising on major search engines, we promote the platform with banner messages displayed on the online social networking service Facebook. As well, we post messages about Pioneering Healthcare on the social networking service Twitter and on LinkedIn using accounts operated by F. Hoffmann-La Roche Ltd.

Visitors to the platform are able to assess their eligibility to participate in Roche-sponsored clinical trials. An online form enables visitors to enter basic details about their diagnosis and treatment history. This information is compared with the eligibility and exclusion criteria corresponding to ongoing Roche-sponsored clinical trials. The details submitted on the platform are not stored, and the online form does not capture information concerning the visitor's identity such as their name or physical location. At the same time, the information we obtain from the form enables us to roughly estimate the proportion of website visitors who would be eligible for enrolment in specific clinical trials. Visitors who complete the form are given feedback about the likelihood that they could participate in a clinical study. The feedback page features the Trial Information Support Line telephone number to enable visitors to seek further information.

In summary, the primary goal of our platform is to inform people about the contribution of clinical trials to the development of better cancer treatment options. The platform hosts a succinct clinical summary of lung cancer and describes processes relating to clinical trial enrollment such as study inclusion and exclusion criteria and informed consent. Importantly, the platform presents the views and responses of individuals who visit the site. While this content may be considered secondary to the goal of informing people about medical and regulatory aspects of the clinical trial process, it provides important contextual information for people as they choose their own way forward following a diagnosis with cancer. To the best of our knowledge, other sponsors of cancer clinical development are not using this approach to boost awareness of clinical trials.

Figure 1. Overview of the Pioneering Healthcare platform highlighting video content, the blog, online polls, and activity on social media. Across the platform, the visual imagery is designed to communicate a sense of community and encourage visitors to engage with others online.

Developing the Platform

The platform has been developed through an innovative collaboration between clinical operations experts and technologists at Roche. The website is based on an industry standard content management system (CMS). So-called plug-ins are used to extend the functionality of the standard CMS, including website integration with social media services and the operation of interactive online forms. A key feature of the platform is the use of digital technologies to engage in two-way interactions. The technologies enable an interaction with other website visitors, and with the operators of the platform itself. This focus on two-way interaction adds to, and complements, our goal to publish and deliver useful and emotionally supportive information to lung cancer patients. The focus on two-way interaction is aligned with the goals of Internet discussion systems characteristic of the pre-website era, such as the World Wide Web discussion system Usenet. Usenet, established in 1979, is still active today and hosts millions of digital conversations between individuals. Usenet's enduring presence is strong confirmation of the need for two-way interaction on the Internet.

In addition to providing information about a particular topic, these systems support people seeking to engage with other people with whom they share a particular interest. Internet-based discussion systems enable people to search for, and make contact with other people. This engagement can be both empowering and rewarding for patients [11,12].

Visitors to the Pioneering Healthcare platform achieve this two-way interaction using online forms. The messages are recorded and sent automatically to Roche via email for the purposes of monitoring and content moderation before it is published on the platform. Visitors can also share links to the platform on social media services such as Facebook. The platform meets US and European regulatory requirements pertaining to patient safety. Comments and other messages received on the platform are monitored for adverse event content. If a visitor to the platform contributes a description of an adverse event then this is reported, within one business day, to the relevant regulatory compliance personnel within the organization. Those monitoring the website are trained in all relevant reporting procedures and follow industry guidelines relating to adverse event reporting.

New content is added continuously to the site in order to stimulate visitor engagement [13,14], and to guide discussion around topics that are relevant to patients and caregivers. We encourage visitors to reach out to other cancer survivors and caregivers, and to take an active role in decision-making their treatment. These messages align with recommendations published by cancer advocacy organizations such as the American Cancer Society and Cancer Research UK. The messages are also in accordance with an active literature examining the connection between information seeking behavior and positive patient perceptions of treatment and treatment outcomes [11,15].

By educating themselves and taking an active role in their care, patients become activated and more involved in their own care and treatment [15]. Self-efficacy and information seeking behavior of someone acting on their own behalf may help an individual to cope with the uncertainties of life-threatening illness [15,16]. We acknowledge that facing advanced cancer is enormously challenging. In according with the academic literature [3,6] we urge visitors to seek professional help if they experience feelings of grief and helplessness.

The resulting activity on the website is tracked and monitored systematically. We track conventional statistics such as the number of times each page is visited, as well as detailed statistics on how visitors move around the website, the pages they share

on social media, and the number of messages and comments they contribute. This analysis is used to continuously refine and optimize the platform.

In developing the platform we have collaborated extensively with the legal and compliance departments at Roche. Medical content is reviewed and approved by clinicians within the organization that have specialized knowledge of relevant disease areas. Reviewers also have a sound knowledge of Roche's ongoing clinical development plans. Collaborations with medical, legal, and compliance experts began during the initial planning phase of the platform, and these collaborations remain active now that the platform functions in an operational mode. The sustained nature of these collaborations ensures ongoing access to expert reviewers. In turn, ongoing access to reviewers ensures that we can maintain operational safety and compliance.

Operating the Platform

The content appearing on the Pioneering Healthcare website is managed continuously, and can be modified instantaneously by a support team within Roche. The website and email server is hosted by a third party service provider offering support on a 24-hour basis 7 days a week and 365 days a year. A digital community manager is deployed to moderate interactions with visitors to the website. The community manager moderates all messages and comments before they appear live on the website. Participants of online communities have defined expectations and norms. Beyond the norms around standards of politeness and consideration, online communities demand rapid and careful management of each and every message that is sent and received on the platform. Close and continuous monitoring by the community manager enables timely and considered responses.

Each new comment or message contributed to the site is first classified according to its suitability for publication. This classification is made in terms of the content of the message, as well as the message's emotional tone. Using email as a means to communicate with an individual contributor, the community manager may propose to publish contributions without modification, redact portions of the content, or alternatively advise the contributor politely that the comment or message is not suitable for publication. Moderation procedures ensure that published messages adhere to community standards of politeness, care, and consideration for our target audience, namely people with advanced cancer. The community manager responds to individual queries and may direct visitors to scientifically reputable online resources such as the US National Institutes of Health National Library of Medicine.

The community manager also adheres to standards of accuracy in the publication of visitor-contributed content, particularly in cases where medical and treatment themes are contributed. Contributions are reviewed and checked against specialised clinical publications maintained by F. Hoffmann-La Roche Ltd, as well as online resources maintained by the US National Library of Medicine. The community manager is also able to call upon medical and legal experts within Roche to perform this task.

We do not publish the names of individual pharmaceutical products on the platform. When a visitor includes a product name in a message or comment, the community manager seeks permission to replace the product name with the name corresponding to the pharmacological class of medications to which the product belongs. Alternatively, the community manager may propose that a specific treatment be redacted from a message or comment published on the website. This option is preferred when a visitor submits a message that makes reference to treatments that may be considered controversial by academic institutions or regulatory organizations.

The website blog is updated frequently and covers practical themes such as healthy diets for cancer survivors, advice about communicating with friends, and about services for lung cancer patients. The blog is also used to publish interviews with health care professionals and others who have expertise in the area of lung cancer treatment and survivorship. Blog posts are made to coincide with events such as the New Year and with changes in the season. These blog posts invite readers to share messages about their experiences at these emotionally important times of the year. Delivering new and topical content at such times can be an especially effective way to support patients and caregivers.

Our readers are kept up-to-date with announcements about major conferences, for example the annual American Society of Clinical Oncology (ASCO) meeting. A relatively small number of annual conferences, including ASCO, make a major contribution to the stock of new and important cancer research insights. Major conferences are, therefore, especially valuable for non-experts who seek accurate and up-to-date information. The organisers of major medical conferences routinely produce materials targeted at lay audiences. These materials include infographics showing important epidemiological statistics, lung cancer pathology, and diagrammatic presentations of a medicine's mode of action.

Rather than duplicate the efforts made by these organisations, we seek to intermediate between patients and the organisers of these events with links to online information intended for lay audiences. The value we add is to direct our visitors to specialized, and up-to-date, information on a continuous basis. These links are carefully selected to ensure that our community is presented with information of a practical nature. As with the content we publish on the website, we distribute links to materials that offer hope and inspiration. We generally avoid materials relating to pre-clinical studies, or to otherwise speculative academic insights that are not yet available in the clinic.

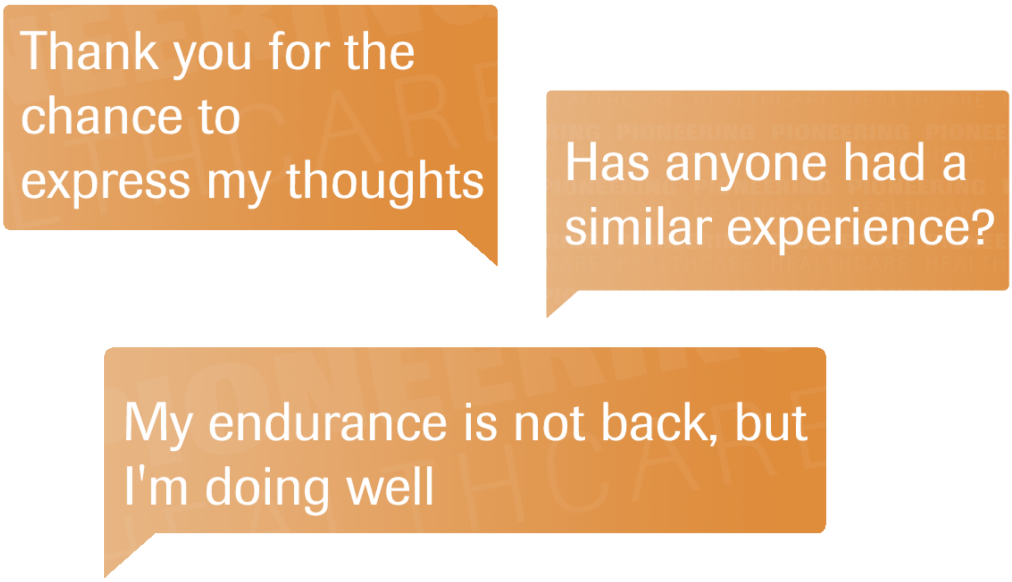
The blog also provides advance notice about important patient-focused conferences, such as the annual Medicine X conference organised by Stanford University School of Medicine. Conferences such as Medicine X, and other conference platforms such as TED (Technology, Entertainment, Design) provide psychological, as well as informational, support for patients and carers. Importantly, we include advice about how to follow these events and conferences using social media. This advice takes the form of practical search tips to be used on platforms such as Twitter and Facebook, and for search engines such as Google. These tips enable visitors to find

relevant content. In addition, these search tips enable our visitors to find people with shared interests and to engage with them online. It is the nature of social media that content is contributed by a large number of individuals. Individuals may contribute links to new research findings and novel insights. Others contribute by sharing this content and adding their own comments and views. Identifying suitable individuals for our visitors to engage with online requires careful research and validation. We direct our visitors to a relatively small number of experts and advocates to ensure that our visitors have a manageable list of individuals to follow and engage with.

The practical advice we provide about searching for content online is undergoing constant change and evolution. We deploy quantitative methods to track and validate the advice we provide to our visitors. By tracking online trends and publishing practical advice about online search we can reduce the effort that our visitors would otherwise need to make. Our visitors can use this information to engage online from the comfort and privacy of their home.

Finally, the blog chronicles the online activity on our own platform. We make blog posts to highlight messages from visitors to the platform. A blog might refer to a visitor who sought further information about a particular topic. The blog post would feature links to where this information can be found online, and acknowledge and thank the visitor for bringing the query to our platform. Special blog posts are made when patient advocates visit the website and leave a comment on the platform. We focus on patient advocates who collaborate with research organisations, or non-profit organisations that sponsor research. In such blog posts we would mention the individual visitor by name, and profile the person briefly using publicly available information about the person's advocacy activities. By mentioning the names of known patient advocates we inform our regular visitors about high profile activities within the lung cancer community. This information is of a high standard, because patient advocates who associate with research organisations and major non-profit organisations are generally vetted carefully by those organisations. The resulting content can be revisited and shared by individuals who have themselves contributed content to the platform. This in itself may be a benefit for patients and caregivers [2]. Visitors to the platform explicitly communicate their appreciation for the opportunity to publish their experiences on our platform (Figure 2).

Figure 2. Excerpts from messages contributed by visitors to the Pioneering Healthcare platform.



Discussion

We deployed the technical platform within 6 months (April 2014), including the time required to complete a review of the website content by corporate representatives from Roche's legal and compliance departments. Rapid deployment to a live website was achieved using a standard online content management system and interactive features have been added using software plug-ins.

We have enjoyed a steady flow of messages and comments from visitors to the platform, as well as a gradual increase in the proportion of return visitors to the website (Figure 3). In most cases biographical information is contributed, focusing especially on the time period from initial diagnosis and treatment. Many messages are from caregivers, including close family members. These messages from visitors to the website have proven to be highly informative in our efforts to adapt and optimize the content we publish on the Pioneering Healthcare platform. The messages we receive confirm that survivors and caregivers seek practical information about diet, their treatment options, and about dealing with feelings of isolation. The messages also confirm the sensitivities around end of life care and the need to maintain a hopeful and positive orientation.

The platform enables us to maintain an informal collaboration with patients, caregivers, and health care professionals. The collaboration is informal in the sense that visitors offer their messages voluntarily. The only restriction applying to the collaboration is that the contributions adhere to the norms of politeness and consideration for other visitors. In return, the messages that visitors contribute are published and maintained online for them. The platform provides visitors with an online

presence, and contributors can share links to their messages and know that others can read their messages and respond to these messages on the platform.

The overwhelming majority of content submitted to the platform comes from patients and caregivers matching the precise diagnosis of non small-cell lung cancer that we seek to enroll in our clinical trial. This may be partly due to dominance of this patient group among lung cancer survivors [17]. It may also suggest that the targeting and promotion of the platform is effective at reaching this precise audience.

We find few people wishing to post overtly negative or even aggressive messages or critical comments about the pharmaceutical industry in general. We respond via email to those visitors and remind them of the purpose of our platform: advancing clinical research. If required, we inform the contributor politely that their content is not suitable for publication on our platform and suggest publishing their opinions on tailored discussion channels.

We use social media platforms (ie, Twitter and Facebook) to follow current trends and discussions relating to lung cancer. This digital listening activity facilitates the generation of new content for the website and enables us to follow important annual online campaigns, including lung cancer awareness month in November and World Cancer Day in February. Digital listening also enables us to have a reliable and up to date understanding of social media mores that are observed by participants in the online conversation about lung cancer. This helps us connect with our audience, and also helps to manage risks to Roche's reputation on digital channels.

Finally, we engage with individuals using social media services such as Twitter and LinkedIn. This engagement takes the form of posts that we publish on Roche's Twitter and LinkedIn accounts. The posts acknowledge individuals by mentioning their name, and commenting on the contribution that they make to the lives of patients and caregivers. Digital listening enables us to select these individuals carefully, avoiding risky online conversations that might damage Roche's reputation. These activities bring us into contact with patients and health care professionals around the world.

Our platform is unique in its focus on emotional support for patients and caregivers, and in the strategy to deliver this support via an online community. A host of digital features including an interactive app, continuous content updates, and representation on social media channels enable us to deliver this support directly and on a continuous basis.

A novelty of our approach is that the platform supports visitors in their own efforts to locate and understand health information online. Practical tips published on the platform enable visitors to search for relevant online content themselves, and to connect with an extended community of people who have shared interests and who are active online.

In summary, the unique focus on emotional support and digital community is yielding authentic interactions with patients and caregivers, and boosting awareness of clinical development in the cancer field.

Figure 3. Percentage of new visitors as a percentage of existing or previous visitors. The initial 10 months of operation are shown. We observe a gradual increase in the percentage of return visitors over time.

Outlook

A new form of collaboration is developing between patients, health care professionals, and the sponsors of clinical trials. Guided by the need to accelerate patient recruitment and make clinical trial enrolment and retention more efficient, the sponsors of drug development are carefully listening to patients' views and motivations.

For patients who are already enrolled in a clinical trial, this new form of collaboration is most clearly manifest in the routine inclusion of patient reported outcomes (PRO) in late stage clinical trials. Formal surveys such as the European Organization for Research and Treatment of Cancer Quality of Life Questionnaire bring a systematic quality to the acquisition and analysis of patient views.

Digital platforms may soon deliver a similar systematic quality to the acquisition and analysis of patient views. Digital platforms enable us to reach a large number of patients, but also extend our reach to caregivers, health care professionals, and other individuals who are active in patient advocacy activities. Interactive websites gather the messages and insights of a global audience, and in real time. The abundant availability of website traffic information, and other data capturing online behavior,

means that this information can be analyzed with significant depth and precision.

In the creation of the platform, we have collaborated with experts in clinical operations and leveraged internal medical expertise at Roche to meet complex safety and compliance requirements. We adapt the platform continuously to ensure that it remains up to date with our current clinical development needs.

Pioneering Healthcare shows what can be achieved when clinical operations experts and technologists work together to host direct interactions with patients and caregivers. The authentic online conversation between clinical trial sponsor and patients on the Pioneering Healthcare platform deepens our insight into the needs of patients with advanced cancer.

The challenge we face is to fully understand all aspects of the insights gained from Pioneering Healthcare's informal online collaborations with patients and caregivers. Importantly, the challenge remains to deploy these insights in the service of those impacted by advanced cancer, and in the development of the next generation of medical treatments.

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Authors' Contributions

The Pioneering Healthcare platform has been developed through a collaboration between clinical operations experts Lea Haines, Shuree Harrison, Margaret Chan, and Mark Wygonik as well as technologists Christian Gossens, Michael Lindemann, Tobe Freeman, and Timothy Kilchenmann. Tobe Freeman, Michael Lindemann, and Christian Gossens authored the paper.

Conflicts of Interest

None declared.

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Abbreviations

ASCO: American Society of Clinical Oncology
CMS: content management system

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Original Paper

The Australian Longitudinal Study on Women's Health: Using Focus Groups to Inform Recruitment

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Abstract

Background: Recruitment and retention of participants to large-scale, longitudinal studies can be a challenge, particularly when trying to target young women. Qualitative inquiries with members of the target population can prove valuable in assisting with the development of effective recruiting techniques. Researchers in the current study made use of focus group methodology to identify how to encourage young women aged 18-23 to participate in a national cohort online survey.

Objective: Our objectives were to gain insight into how to encourage young women to participate in a large-scale, longitudinal health survey, as well as to evaluate the survey instrument and mode of administration.

Methods: The Australian Longitudinal Study on Women's Health used focus group methodology to learn how to encourage young women to participate in a large-scale, longitudinal Web-based health survey and to evaluate the survey instrument and mode of administration. Nineteen groups, involving 75 women aged 18-23 years, were held in remote, regional, and urban areas of New South Wales and Queensland.

Results: Focus groups were held in 2 stages, with discussions lasting from 19 minutes to over 1 hour. The focus groups allowed concord to be reached regarding survey promotion using social media, why personal information was needed, strategies to ensure confidentiality, how best to ask sensitive questions, and survey design for ease of completion. Recruitment into the focus groups proved difficult: the groups varied in size between 1 and 8 participants, with the majority conducted with 2 participants.

Conclusions: Intense recruitment efforts and variation in final focus group numbers highlights the "hard to reach" character of young women. However, the benefits of conducting focus group discussions as a preparatory stage to the recruitment of a large cohort for a longitudinal Web-based health survey were upheld.

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KEYWORDS

Focus groups; methodology; surveys; longitudinal studies; participant recruitment; social media; web-based survey; mobile phones

Introduction

Recruiting and retention of participants to large-scale longitudinal studies has become a challenge in recent years [1], particularly when resources are limited and low response rates lead to the costs of more traditional random sampling methods becoming prohibitive [2]. Qualitative inquiries with members of the target population can assist in the development of effective recruiting techniques [3]. The current study utilized qualitative methods to examine the facilitators and barriers to

participation in a national cohort online survey study of women aged 18-23 years.

Existing qualitative research has indicated a number of factors that encourage participation in health research and how to maximize recruitment. Demonstrating the importance of remaining alert during the recruitment phase, Dyas et al reported upon recruitment to a focus group study as part of a primary care sleep intervention in the United Kingdom [3]. While monitoring the recruitment of participants into the focus groups, the authors noted evidence of recruitment tactics not in keeping with the original research plan, and a number of changes were necessary to their original recruitment strategy to boost lagging participation numbers. In their findings, the authors reiterated the importance of only using evidence of what works best in the planning and design of studies. In terms of recruitment of participants into health research, Dyas et al noted that although individuals can be interested in a topic it does not necessarily equate to subsequent participation [3]. This point was supported by de Jonge [4] when referring to recruitment into focus groups specifically. The author found that due to recruitment difficulties during her work on support for teenage mothers, she was unable to conduct focus groups as originally planned, which led to fewer useful comments from study participants.

Recruiting young women into research can present unique challenges [5-6], with poor response rates a common problem [7-8] and limited details available in the literature on methods for establishing and recruiting young women for longitudinal studies [9]. For the current study to identify how to encourage women aged 18-23 to participate in a national cohort online survey, researchers had to learn more about the challenges involved from young women themselves. Focus groups can provide researchers with a means of listening to the perspective of their target study participants, as well as an opportunity to learn from participants' opinions and experiences about participation in health research [10]. For example, Herbert, Loxton, Bateson, Weisberg, and Lucke [2] held 10 focus group discussions with young women in preparation for their Internet-based study on contraceptive use and pregnancy intentions. Findings indicated that participants held concerns about the confidentiality of the Internet, were open to being asked about sensitive topics, favored incentives, and wanted an attractive survey with relevant content. In contrast, Giles, Sniehotta, Mccoll, and Adams [11] grouped together participants whose ages ranged from 18 to 59 years in their group discussions to explore the acceptability of incentives to influence healthier behaviors. This broad mix of life stages and experiences may have led to some participants not feeling comfortable explaining their unhealthy behaviors in front of others.

As demonstrated by Herbert et al, focus group methodology can be an appropriate form of inquiry when investigating effective recruitment strategies. Brought to prominence over time by market researchers, the focus group method allows people to explore and clarify views on particular topics within a group environment [12] and in ways more easily accessible compared to a 1-on-1 interview. Of particular bearing to the current study with women aged 18-23 years, is that focus groups can encourage research participants to explore issues that are important to them, in their own vocabulary [13].

Focus groups proved useful for Brown et al [14] who examined job satisfaction of research recruiters working at academic research institutions and health maintenance organizations tasked with recruitment of diverse groups of women into research, although 27 participants were split into only 2 focus groups, which could mean that many women were not given an equal opportunity to be heard. A large-scale cohort study in NSW Australia that examined risk factors for injury in drivers aged 17-24 used focus groups to discuss modification of recruitment techniques; however, they were not reported in detail and therefore little is known about their use [15].

Work by Ungar et al [16] into early prevention of aggression in children used focus groups to help probe quantitative outcome data 4 years after study completion. The authors reported that their qualitative evaluation methodologies resulted in some respondent bias (ie, those who agreed to participate 4 years later) and recall bias (ie, problems remembering the original intervention). Novel work by Chatrakul Na Ayudhya et al [17] applied a life course perspective through focus group methodology to explore young adult experiences of transitioning from university to full-time employment. However, the authors did not provide details regarding participant recruitment, which would have provided greater context to their results.

Overall, despite the widespread use of focus groups in social and health research, there are few detailed accounts of using focus group methods to inform and customize recruitment approaches for a large-scale health study in Australia, particularly one that requires participation at a national level for an indeterminate amount of time. The current study sought to add to existing knowledge by assessing women's willingness to sign up for a study that has no effective end date (ie, is longitudinal). Longitudinal studies such as this can last a lifetime, and engaging young people into such long-term projects is challenging. Further, this current study will endeavor to remove uncertainties about how focus groups are organized and conducted, through transparency and level of detail reported.

This study firstly aimed to gain insight into how to encourage young women to participate in a large-scale, longitudinal health survey. Secondly it aimed to evaluate the survey instrument and mode of administration. Using focus group methodology with young women aged 18-23 years old from remote, regional, and urban areas of 2 Australian states, the following questions would be investigated:

1. What steps could the research team take to attract the attention of young women and encourage their participation in a longitudinal health survey?
2. How could potentially sensitive items be presented?
3. Were there concerns about the provision of personal contact details and, if so, what were these and how could they be addressed?
4. What design features would be likely to assist young women to complete a health survey?

Methods

Study Context

This paper describes one aspect of preparations by researchers from the Australian Longitudinal Study on Women's Health (ALSWH) to recruit a new cohort of women born during 1989-95. ALSWH is a multidisciplinary project that conducted baseline surveys with 41,449 women across 3 birth cohorts in 1996 and continues to survey participants on a rolling basis every 3 years [18,19], with the 1921-26 cohort receiving surveys every 6 months as of 2011. The 1989-95 cohort represents a new generation of Australian women being recruited in a different era—18 years after the original ALSWH baseline surveys—and provides researchers with the challenge of adapting the existing ALSWH survey methodology to suit this generation's lifestyle preferences and needs. The original study sample for ALSWH was selected by Medicare Australia (previously known as the Health Insurance Commission) from a database containing the name and address details of all Australian citizens and permanent residents [20]. Women were randomly sampled and mailed an introductory letter, information brochure, consent form, a paper copy of the survey, and a reply paid envelope, followed by a series of reminders [20].

The potential success of recruiting women aged 18-23 years via Medicare was gauged by researchers through observations of recent research by Herbert et al [2]. Of the 900 women sent a written paper invitation by Medicare to participate in the CUPID project's Web-based survey, only 47 women (5%) completed the survey. This rose to 51 responses (6%) after reminder letters were sent out, but was significantly lower than the 41% response rate (14,792 women out of 36,067) experienced by ALSWH using this method in 1996 [18]. Clearly a different approach had to be explored for a new generation.

Focus Group Processes

Focus groups can prove valuable during the formative stages of research and were deemed the most appropriate method through which to elucidate young women's points of view, needs, and concerns regarding being invited to complete a health survey as part of ALSWH [21]. The aim is to achieve general agreement from young women regarding their participation in a health survey. The focus group moderators were both female: the first was a 22-year-old ALSWH employee with experience conducting focus groups with young women, and the second was a 36-year-old psychology graduate with experience in conducting qualitative and quantitative research.

The focus groups used a semi-structured interview guide to encourage discussion of specific survey-related topics while offering flexibility of conversation between group members. Selection criteria for the focus groups required participants to be female, aged 18-23 years old, living in Australia, willing to volunteer to participate in a focus group discussion, and proficient in the English language. To encourage a wider range of potential opinions, diversity of participation was sought. Selected Australian Bureau of Statistics "snapshot reports" were used to identify focus group locations based on demographic, geographic, socioeconomic, and cultural characteristics. Within these demographic parameters, considerations were also made in terms of travel expense and whether ALSWH researchers had contacts that could generate interest around the areas where the focus groups were being held.

Fourteen focus groups were planned, with approximately 5-10 participants in each (minimum 70, maximum 140 participants). The focus groups were organized in 2 stages: the first to examine how to encourage young women to participate in a large-scale, longitudinal health survey (study aim 1), and the second to evaluate the survey instrument and mode of administration (study aim 2). Groups were planned for remote, regional, and urban settings of New South Wales and Queensland, conducted in community meeting and conference rooms. Participants were recruited via posters placed in prominent areas within the community of a focus group site (eg, libraries, universities, and technical and further education (TAFE) campuses) and via interested community groups (eg, hair salons and fitness centers). The technique of "snowball sampling" [22] was also used after Herbert et al found it to be successful in recruiting young women as focus groups participants [2].

Participants were required to read a detailed information statement and were provided with informed consent forms prior to the start of each focus group. A 7-item written questionnaire was used to collect information on demographic characteristics (eg, age, occupation, educational qualifications, financial situation, work status, student status) and access to the Internet (eg, type of online device used, frequency of Internet use). The first series of focus groups planned to canvass the opinions of young women on issues of methodology, such as: the format, appearance, mode of administration (ie, specifically whether a Web- or paper-based survey was preferred), and promotion of the survey so as to best appeal to the target population. The groups were also used to identify potential concerns about the survey that would need to be addressed, particularly in regard to privacy, confidentiality, and data linkage. Findings from these first groups would be used to draft survey materials, which were then used in later focus groups to examine their acceptability and utility.

Data Analysis

Each focus group was audio recorded and transcribed verbatim. While participants in each focus group were instructed not to use their names as they spoke, any unintended personal information that remained after transcription was removed by the moderators who checked through each transcript and compared them against their own memos.

Analysis was conducted by 2 researchers (RM, MT) with expertise analyzing qualitative data and working with ALSWH data. Neither had been involved in the organization or conduct of the focus groups, and were less likely to be influenced by personal impressions of the participants or potentially allow the results to be affected by a single focus group or participant voice. NVivo software (V10; QSR International, Doncaster, Victoria, Australia) was used to aid thematic analysis, guided by Bazeley's work for coding to single and multiple nodes [23,24]. Firstly, each printed transcript was systematically read and reread with annotations added by hand where necessary. Focus group data were analyzed both deductively and inductively [25]. Anticipated subjects, often introduced to the

groups by the focus group moderator (such as through the use of Facebook to recruit participants and concerns about the length of the survey) were treated as topic-based codes. Subjects that were identified during the analyses (such as focus group participants' emphasis on the importance of explaining why the research was being conducted) were treated as analytic codes.

Focus groups conducted at Stage 1 were transcribed and analyzed as 1 group for the purposes of informing the Web-based survey to be pretested by Stage 2 focus group participants. Answers received during the pretesting of the draft survey were not kept by the researchers, but were instead used as a point of discussion at the time of each focus group to evaluate the survey process and content. Results from a demographic questionnaire completed by all focus group participants were used to describe the focus group sample, ensure diversity, and determine young women's use of the Internet to inform the Web-based survey.

Ethical approval was provided for the conduct of focus groups by the Universities of Newcastle and Queensland. In accordance with ethics committee guidelines, participants were provided with a verbal summary after each focus group by the moderators, based on their notes. Other than consenting participants and the moderators, no other person was present during the discussions.

Standards of Rigor

This manuscript was guided by a recent review of focus group studies by Carlsen and Glenton [26] who conducted a review of 220 studies that had used focus groups and reported that focus group methodology was often poorly described. As such, this study adopts careful reporting of the number and size of focus groups conducted and lessons learned regarding use of focus groups to inform large-scale health surveys. The criteria by Tong and colleagues [27] for reporting qualitative research involving interviews or focus groups is also acknowledged in reporting the conduct of this study.

Results

Study Sample

A total of 19 focus groups were held, in 2 stages from September 12, 2011, to April 12, 2012. For Stage 1 discussions regarding how to encourage young women to participate in a large-scale, longitudinal health survey, 13 groups involving 56 participants were conducted in 5 locations throughout New South Wales (NSW) and Queensland (Qld). The time taken ranged from 19 minutes to 1 hour 8 minutes. For Stage 2 where a draft survey was pretested, 6 group discussions involving 19 participants were conducted in 3 suburbs in a regional city of NSW. The time taken ranged from 44 minutes to 1 hour 6 minutes. The groups varied in size between 1 (1 focus group only) and 8 participants (2 focus groups). The majority of focus groups were conducted with 2 participants (5 groups). The majority of participants were 20 (21/75, 28%) to 21 years of age (16/75, 22%), held or were completing a university degree (36/75, 48%), and lived in a regional area (49/75, 65%). Summary demographic information for all participants is shown in Table 1.

Analysis of Focus Group Discussions

Thematic analyses of the 19 focus group transcripts identified several primary themes, which guide our findings as follows:

1. Attracting attention and encouraging participation;

2. Survey length, presentation, and administration;

3. Survey content, including potentially sensitive questions; and

4. Providing personal details and follow-up.

A summary of the themes, their definitions and key examples of participant discussions are provided in Table 2.

Table 1. The demographic profiles of focus group participants.

Demographic variable		No. of participants (N=75)	% of total
Age, y			
	18	13	17%
	19	10	13%
	20	21	28%
	21	16	22%
	22	5	7%
	23	10	13%
Area of residence			
	Major regional city, NSW	31	41%
	Outer major city suburb, NSW	6	8%
	Major city, Qld	11	15%
	Inland very remote town, Qld	9	12%
	Inland regional town, Qld	18	24%
Highest educational qualification			
	Year 10	3	4%
	Year 11	2	3%
	Year 12	28	37%
	TAFE/Vocational	6	8%
	Held or completing a university degree	36	48%
Work status			
	Full-time work	10	13%
	Part-time work	13	17%
	Casual work	36	48%
	Not working	16	22%
Student status			
	Full-time study	56	75%
	Part-time study	4	5%
	Not studying	15	20%
How do you manage on your income?			
	It is impossible	2	3%
	It is difficult all the time	6	8%
	It is difficult some of the time	27	36%
	It is not too bad	29	39%
	It is easy	10	13%
	Missing data	1	1%

Table 2. Summary of focus group themes and definitions.

Theme	Definition	Key quote examples from Results section
Attracting attention and encouraging participation	Ways to get the attention of young women, whether (and how) to use social media to best effect, and how researchers could explain participation and benefits of a health study	"A lot of the things that I sort of see, whether it's for charity or fundraisers, that sort of thing, is always through Facebook."
Survey length, presentation, and administration	Survey design ideas and ways to facilitate completion of an online survey by young women	"I find things on the Internet, if I get sent a link and all I have to do is click on it, then I'm happy to do it."
Survey content, including potentially sensitive questions	Why some questions were included, how to phrase questions considered "sensitive," and layout for electronic devices	"Maybe having the option of choosing not to answer it as well...That's probably better than making you answer and not answering truthfully for things."
Providing personal details and follow-up	How best to legitimatize the study to participants, fears about the confidentiality of information, concerns regarding providing personal information, and permission for data linkage	"I would be happy to put my phone number, my home address, but I wouldn't want to put that with my date of birth 'cause, I don't know, my dad's all paranoid about, like, identity theft."

Attracting Attention and Encouraging Participation

Key strategies discussed by focus group participants for engaging young women were promoting the survey on social media, explaining why their information was needed, and offering financial incentives for survey completion.

Overwhelmingly, participants stated that social media (particularly Facebook) was important for connecting with the 18- to 23-year-old age group, as a participant said, "It can reach a lot of people." Facebook was highlighted in 12 out of 13 Stage 1 focus groups as a positive or important strategy for promoting the research project and the survey. Overall, however, participants said that television "is pretty much out, because we don't watch TV." This was particularly true for women who lived in a university student residence where TV was not accessed as much. Other publicity methods suggested by participants included handing out flyers at music festivals and universities, advertising on radio, and placing articles in newspapers and women's magazines, such as Cosmopolitan and Cleo:

> *Because I live in a share house with 4 girls and we always have Cosmo and all of that in there...obviously it's a women's magazine, it's going to be appropriate...That could maybe be a good way of getting to who you want to get to...[your] target market I suppose.* [Group 6, major city, Qld]

Participants in Stage 2 focus groups were asked to suggest alternative options to Facebook if the ALSWH could not have a Facebook presence. Discussions in 4 out of 6 focus groups still featured Facebook advertisements as an alternative strategy to a project-specific Facebook page, which highlights the importance of Facebook as a way to connect to this age group. As 1 woman asked, "Is there a world outside Facebook?" [Group 19, major regional city, NSW]

A strong preference was also expressed for a link to the survey from social media sites and postings:

> *A lot of the things that I sort of see, whether it's for charity or fundraisers, that sort of thing, is always through Facebook. Either a page or an event [with] links to other places from there.* [Group 6, major city, Qld]

However, placing advertisement links for the survey in the side-column of Facebook was seen as a potential virus source and lacking in legitimacy. In this instance, authenticity would be increased by embedding the health survey within the wider ALSWH "brand." Additionally, using a study, university, or government logo was discussed by 5 groups during Stage 1 as a strategy to help minimize potential suspicion and unease.

According to participants from 16 of the 19 focus groups from both Stage 1 and 2, part of the promotion and advertising initiative for the survey needed to involve information sharing about the purpose and processes of the research. Emphasizing the age-specific relevance and longitudinal nature of the research and the significance and potential impact of participation would make young women want to take part through a feeling of ownership, as only they could provide the information. Giving them a sense of importance and worth, as well as providing them with an understanding of the reason behind the survey and the included questions, was deemed important:

> *You feel almost special because you're in that age group and there's not that probably many people in that age group, so...what you get from it really means something, and that it's really targeted to you, your age, your friends, and all that sort of stuff.* [Group 9, inland very remote town, Qld]

An approach that focused on the "value of research" and the sharing of the research findings was favored by many of the focus group participants' as a strategy that would appeal to them personally, and also to those women who could appreciate the research process. This perspective was held by the 2 participants of 1 particular focus group [Group 4, major city, Qld] who described themselves as having a research background and recognized the "long-term benefits" of research and how "difficult" it was to conduct. It was recognized, however, that others who did not share their background would not necessarily feel like this. For example, 1 participant in another group stated that her participation in the survey would "depend on how bored I was and if it was pretty or not."

Several women stated that they would complete the survey "if I got free stuff." In terms of offering incentives for survey participation, each group in Stage 1 saw it as beneficial to offer the chance to win a "prize," such as coffee, fuel, a movie, or shopping center vouchers, particularly if the survey was long. A number of the ideas were in keeping with a health survey, including a free consultation with a general practitioner. Keeping in mind women in more rural areas, some focus group participants suggested that vouchers or prizes be accessible for all, not just for women living in urban areas. Participants noted that prizes need to be relevant to the 18- to 23-year-old age group, since if it was "just about winning...you just want to win something." [Group 18, major regional city, NSW]

Survey Length, Presentation, and Administration

Of particular concern to participants was the length of time needed to complete the survey. The acceptable time frames articulated by Stage 1 focus group participants ranged from only a few minutes to 20 minutes, although 1 participant stated "however long it takes to finish." When participants were asked how they would feel if the survey could take up to 45 minutes to complete, only a small number said they would continue (those who identified they worked or studied in health services), while most participants felt it was too long, as young people are so "busy" and some "have a very short attention span." Another concern about survey length was linked to completing the survey on a smartphone due to data usage restrictions.

The women suggested survey design ideas that would offset the time taken to complete the survey, including a visual progress bar if the survey was online and adding some color and possibly pop-up information boxes. At the same time, the survey had to look "professional," simple, and not "vile" or "childish," and should not include advertisements. Images were favored as long as they did not detract from the page loading speed and were relevant to women's health. Focus group participants suggested that, in general, Australian women at the younger end of the 18- to 23-year-old age range would probably prefer bright colors and would want the survey to be "pretty," with a few women being specific enough to suggest dusky pink as an appropriate color for a women's health survey.

Those women who participated in the second stage of focus groups were asked to pretest a timed draft survey using a variety of media. The survey took between 8 and 34 minutes to complete. Using a computer was the fastest method (average speed: 15 minutes), followed by using an iPad (17 minutes), and completing the survey by hand (19 minutes). Using a smartphone to complete the survey took the longest (27 minutes). When asked to evaluate the length of time it took them to complete the survey, the majority of participants (18 out of 19) indicated that it was "just right," and 1 participant felt it was "too short."

All participants said they had access to the Internet via computer, with over half (47 out of 75) also using a smartphone to log on. "Other" devices, including tablets, were used by 6 out of 75 participants. Most women described using their mobile phone for specific "on the spot" Internet activities such as checking email, locating maps, online banking, and Facebook. For "more complicated things" such as surveys, they used a computer due to the larger screen and keypad, as well as data download restrictions on their phones. However, it would be necessary for the online Web-based survey to be in a format adaptable for use on a smartphone for those who wanted to utilize this method:

> *It's got to be phone friendly. I think people would be even more inclined to do it if they could do it while they're lying in bed at 11 o'clock at night like I do.* [Group 9, inland very remote town, Qld]

The main difficulty for those completing the draft Web-based survey was typing the URL for the survey from the paper invitation into the Internet address bar. The length and case-sensitive nature of the URL was particularly frustrating for women completing the survey using the touch screen on an iPad or smartphone. It took 1 participant over 6 minutes to access the survey because of this. Focus group participants felt that recruitment via a written postal invitation to complete a Web-based survey would hinder participation due to the delay between reading the paper letter and logging on to the Internet to complete the survey. Stage 2 participants reiterated the preferences of Stage 1 women, advocating the need to be able to immediately "click the link" from an email invitation or online advertisement through to a Web-based survey, removing the possibility of nonparticipation due to frustration, forgetting about the survey, or losing the survey's paper invitation with the Web address:

> *It makes it easier...[if you get]...an email with a link page, you can just control-click and then it brings it up in a new tab....I'm not going to type all those things into the address search bar and then do it, [the URL] would have to be a short thing.* [Group 2, major regional city, NSW]

> *I find things on the Internet, if I get sent a link and all I have to do is click on it then I'm happy to do it. But if I have to go and look it up I kind of either don't remember or I can't be bothered.* [Group 6, major city, Qld]

The appropriateness of the Internet as the principle mode of survey recruitment and administration was supported by the findings from both the focus group discussions and the written demographic surveys, in that almost every focus group participant (71 out of 75) accessed the Internet daily, and often several times a day. The remaining 4 women stated that they used the Internet weekly. The Internet was described as a part of everyday life for the majority of participants:

> *I check it [the Internet] every morning as soon as I wake up and have my breakfast, Facebook, email...every morning...* [Group 1, major regional city, NSW]

Survey Content, Including Potentially Sensitive Questions

Survey content was largely viewed from a practical perspective by participants. This included clarity as to why particular questions were being asked, and having the survey operate efficiently on their chosen electronic device. Features such as offering multiple choice questions, organizing the questions by topic, and asking only a few questions per page were popular

suggestions. Participants suggested that the instructions be clear but brief and for any introductory wording before potentially sensitive questions to be obvious. Also, participants in 9 of the 13 Stage 1 focus groups discussed not liking questions that required "more thought," such as having to calculate time periods.

In keeping with their reported high usage of the Internet, participants said the survey should be formatted for computer, smartphone, and iPad. Primarily they recommended that the survey be easy to read, with large enough font and black text, and should fit well on the screen without having to scroll up and down and left and right to view questions. Long paragraphs of text should be avoided and the text should use laymen's terminology:

> *As long as it's easy to read and it fits on the screen without having to scroll like across and everywhere and scrolling down this massive thing. I think it's probably better just in terms of ease of filling it out.* [Group 3, outer major city suburb, NSW]

Views on response options were mixed, with some women preferring multiple choice questions while others wanted more flexible options to enable them to answer as close as possible to their own situation, such as including a text box for longer answers. Some women preferred scales and liked the option of neutral responses as opposed to forced statements, while women in several groups referred specifically to how much they did not like Likert scales. Every group in Stage 1 mentioned that they did not like repetitive questions, and 16 out of 19 groups across both focus group stages discussed the importance of including an option to "skip" certain questions.

The focus group participants were asked to comment on the inclusion of sensitive questions in the survey, including questions on: health behaviors, such as drug and alcohol use; reproductive health and sex; and questions about traumatic events. Overall, there was consensus that as long as an explanation was provided to clarify why certain questions were being asked, that participant confidentiality was assured, and participants were able to skip the questions if preferred, young women of this age group were unlikely to be offended:

> *You're not too worried about answering stuff like that because you know it's confidential. Maybe more personal things like tragic events and stuff, probably—for some people [it] would be traumatic thinking about it, but yeah, if they don't want to answer then as long as they have that option.* [Group 4, major city, Qld]

Questions involving sex, contraception, and alcohol were the least concerning for participants. Issues of confidentiality were highlighted by the focus group participants with regard to the drug use questions. They were also concerned that survey participants who had experienced traumatic events would find answering questions about them confronting and upsetting. The option to not comment or skip the question was seen as important, as well as the provision of a text box that allows participants to elaborate if they want to. In contrast, some women felt the brevity of a multiple choice response could lessen the upset of a sensitive question. Some participants felt that this would assist with such questions being answered truthfully, while others sometimes expressed uncertainty about whether they themselves would answer particular questions accurately:

> *I think you probably could put in sort of any questions you want but it doesn't necessarily mean people are going to be truthful or answer it....maybe having the option of choosing not to answer it as well...that's probably better than making you answer and not answering truthfully for things.* [Group 3, outer major city suburb, NSW]

Providing Personal Details and Follow-Up

Knowing the study was legitimate and fears of confidentiality were among the main concerns of focus group participants. Despite support for a Web-based survey, participant concerns were linked to the study being online, with wariness of "spam" and "junk mail" and "identity theft." One woman was even concerned about the location of the server where the data would be held:

> *I'd want to know where the server was located wherever my survey results were going to, 'cause I know in particular if it is in the USA then under the Patriot Act that can be accessed by US government and things like that so I would like to know where the server [was]...so that I know where my data is going.* [Group 2, major regional city, NSW]

The focus group participants felt some concerns could be countered by receiving a clear description of the study's purpose, confidentiality procedures, and the reasons behind the questions asked, such as the need for contact details and about the significance of, and processes around, data linkage. Conversely, the Internet was described as a tool that women could use themselves to investigate the study behind the survey (ie, ALSWH) before agreeing to complete it.

Lack of anonymity was seen as a potential deterrent to participation. The feedback from participants in each group regarding privacy and confidentiality showed that it was imperative to include explicit explanations about why personal details are required and information about where personal data will be stored. Participants said they would feel better if all correspondence had the affiliated university logos clearly represented, and that the history of the study should be conveyed in an interesting and succinct way to confirm its authenticity.

Some confusion existed in regard to the longitudinal nature of the survey and the associated necessity to collect contact details to assist follow-up and the women's date of birth to help confirm their identity on future surveys. Similarly, a few participants mistook assurances of confidentiality for anonymity, illustrated by the following quotes:

FG Participant: *Well why would you put your name on it? You wouldn't because it would be like a confidential survey, they'd just—they wouldn't want to know who you are, personally, they just want your information.*

Facilitator: *We would have to know personal details like your name and phone number and age and date of birth and things*

like that because it's a longitudinal study, which means that we'll be surveying the same people over a long period of time.

FG Participant: *Oh shit. The only thing I don't put [on surveys] is date of birth. Like I would be happy to put my phone number, my home address, but I wouldn't want to put that with my date of birth 'cause, I don't know, my dad's all paranoid about, like, identity theft and, like, you only need a few things and you can, like, you know, steal a person's identity.* [Group 10, inland very remote town, Qld]

The 1989-95 cohort was asked about consenting to having their ALSWH survey data linked to their service use data from Medicare, a practice that has been successfully implemented with the original ALSWH cohorts [28]. The overwhelming majority of focus group participants were positive about data linkage between the survey and Medicare, stating that they themselves would consent to this if asked. They did feel, however, that other women, not having the benefit of additional explanations from the focus groups, could question the need for the data linkage and personal information requested and be fearful of identity theft. It was viewed as essential that the process be clearly explained to potential participants, emphasizing the importance and benefit of data linkage and that only service provider use data would be accessed, not diagnoses or other personal information, and reiterating the confidentiality procedures that would be in place. One woman actually felt the connection with Medicare increased the legitimacy of the study:

> *I feel like it almost, it makes it more legitimate, like I'd be almost more inclined to do it because I know Medicare...you know it's serious.* [Group 3, outer major city suburb, NSW]

There was a general consensus that the survey participants also needed the ability to "opt out" of data linkage, regardless of providing information and reassurances about the linkage procedures, otherwise women may choose not to do any part of the survey purely because of the linkage request.

The need for participants to enter their Medicare number in the survey may also be problematic from a practical perspective for this age group of women. Twenty focus group participants indicated that they were still on their parents' Medicare card. Five of these women had their own card but were still linked to the family Medicare number and the remaining 15 would need to phone a parent to ask for the number, delaying and possibly derailing their survey completion.

> *That would make me quit the study as well. If my dad's not home then I'd have to get up and try and call him [to get my Medicare number]. There's no way I'd go back to it [the survey]. Once I start something and I don't finish it, I'm not starting it again.* [Group 18, major regional city, NSW]

The focus group participants were asked to discuss reminder and retention methods that would be put in place after women had been recruited to the study. Two reminders asking participants to complete their survey were considered appropriate, with an email and/or a short message service (SMS) text sent to a mobile phone preferred.

> *Any more than 2—if I need 2, I'm not going to do it. If I haven't done it already and I've had 2 reminders, it's not going to happen.* [Group 4, major city, Qld.]

Contacting participants via phone call or Facebook was considered "too personal and in your face." Using 2 different methods for the reminders was recommended by many of the participants, the reasons given included: not being able to receive one type of contact due to environmental mischances and remoteness. One woman said "...like with floods, too, when we had our floods here, no, we never got mail for a month. We still had Internet access but we didn't have our mail." [Group 12, inland regional town, Qld] Another woman from the same focus group agreed, "Yeah, 2 different methods because some people, they're rural. They wouldn't have frequent access to Internet so mail would probably be the best way for them." [Group 12, inland regional town, Qld]

The email or SMS would need to stand out and each be followed up by the other method as a means of reinforcing the reminder message. Two groups suggested giving participants the option of choosing how they would like to be reminded when they completed the first survey. Some women conceded that more than 2 reminders may be necessary but that if the women had already joined the study, more reminders would be acceptable. Generally, there was consensus that a maximum of 2 reminders between the initial invitation and the survey closing date was acceptable. If more than 2 reminders were required, it was unlikely that the participant was going to complete the survey.

Discussion

Principal Findings

The primary aim—a further understanding of how to encourage young women to participate in a large-scale, longitudinal health survey by using focus group methodology—was met. Group discussions with 75 young women aged 18-23 years old allowed ALSWH researchers to "test the waters" regarding how best to encourage participants' interest, and continued participation in, a health study. Nineteen focus groups were conducted in 2 stages and over a 7-month period across NSW and Qld, Australia. From the 75 women participating, 17 (23%) lived in or near a capital city, 49 (65%) lived in a regional area, and 9 (12%) were in a remote area. The majority of participants were aged 20 or 21 years of age and were in full-time study and/or casual employment. The women had a primary preference for survey promotion via social media and their main concerns regarded giving of personal information, how confidentiality could be assured, and that the health survey be easy and brief to complete.

Comparison with Prior Work

Given the increasing popularity in Web-based surveys and participant recruitment via social media/networking sites ALSWH researchers needed to explore taking a Web-based approach over paper surveys [29-31]. Most young people are adept at using new technologies and are more likely to respond to a Web-based survey than they are to a questionnaire received by post [32]. In terms of how to attract the attention of young women to participate in the ALSWH survey, focus group participants favored social media, email, and SMS text

messaging as tools to connect with the study, for recruitment as well as follow-up. Recent work by Fenner et al showed that social network sites were an effective strategy to use with 16- to 25-year-old Australian females when recruiting people for health research. Particularly of relevance to the current study was the respondents' age distribution, with 18- to 25-year-olds more likely to enroll in the study through social media than 16- to 17-year-olds [33].

The focus group findings supported a Web-based survey as being the most preferred and practical way in which to conduct a large-scale survey. To aid participation by young women in ALSWH the survey had to be designed with convenience, speed, ease, and likelihood of completion in mind. For the participants, the primary advantage of a Web-based survey was that it would take up less of their time. For the researchers this could also mean a higher response rate in a shorter period of time; a finding supported by Leonard et al [5] who compared different recruitment strategies for 18- to 35-year-old women and found that social media was the most successful way to recruit study participants and that using an online survey was the quickest way to secure respondents.

Building a sense of ownership of a study or project can help to build commitment in those taking part and can increase the sustainability of the work [34,35]. In terms of the ALSWH recruitment of a new cohort of women aged 18-23 years old, researchers should allow the women to feel involved by sharing how the health survey is conducted and communicating some study findings. This could improve women's motivation to participate, emphasize the age-specific and longitudinal nature of the research, and help to build their capacity as contributors to knowledge about women's health.

The second research question for the current study asked how potentially sensitive items could be presented in the ALSWH survey. When asked to comment on the inclusion of survey questions asking about drug and alcohol use, reproductive health, and traumatic events, focus group participants acknowledged that while such questions should be answered truthfully, they were uncertain whether they themselves would do so. In order for ALSWH to explain the impact of women's diverse social circumstances on health, the longitudinal health survey needs to obtain accurate data regarding young women's experiences. Although sensitive survey questions can produce higher nonresponse rates [36], respondents will not necessarily withdraw their participation when they encounter sensitive items [37]. Women are also more likely to have far fewer missing answers on highly sensitive questions when the survey is Web-based, compared with men [36,38].

Focus group methodology was also employed by Herbert et al during their research into young women's contraceptive use and pregnancy intentions. They reported that where sensitive items were included in a survey, it was imperative to offer respondents the option to "prefer not to answer" [2]. This is in keeping with Tourangeau and Yan who state that using an appropriate range of response options can help to "avoid forced responses that create false or blank reporting" [39]. Further, focus group participants in the current study agreed that as long as ALSWH provided clarification as to why certain questions were being asked, young women were unlikely to be offended.

In response to whether asking for personal contact details and being sent follow-up reminders would be barriers to participation, focus group participants were definitely hesitant. However, alleviating suspicions regarding privacy and confidentiality could be facilitated by the health survey providing detailed information as to why the research was being conducted and how the research process worked. Attrition is a major concern in longitudinal studies [40]; therefore, the ALSWH health survey for women born 1989-95 will ask for personal address information, the woman's Medicare card number, and consent to link survey answers to other health and administrative databases. Having the Medicare card number will allow the study to verify the participant's details and ensure that the survey was completed by someone of appropriate gender and age. Personal contact details help researchers follow up with the participant for subsequent surveys. Examples of attrition in longitudinal studies reflect the importance of obtaining thorough contact details for participants at the time of the first survey; for example, 45% (4663 out of 10,264) of participants dropped out over 14 survey waves of the British Household Panel Survey [41] and 24% (238 out of 994) were lost over 9 years for the 30-year Finland study of a perinatal birth risk cohort [40].

Practical Implications

ALSWH set out to recruit a new cohort of young women aged 18-23 years old from across Australia. The focus group findings supported the use of nontraditional approaches for recruitment. In turn this led to the design of the ALSWH 1989-95 cohort recruitment strategy, which resulted in the recruitment of over 17,069 participants—16,159 (95%) via social media, targeted online advertising and Web activities, referrals, and incentives, and 910 (5%) via traditional media [42]. Recruited participants were broadly representative of similarly aged young women across Australia [43] in terms of geographical and age distribution, with 95% never married (16,321 out of 17,069) and a majority attaining university (22%, 3844 out of 17,069) and trade/certificate/diploma qualifications (25%, 4428 out of 17,069).

In terms of using focus group methodology to inform research, the groups can be difficult to organize [44], with nonattendance and cancelation all too common. However, the current study found that any challenges experienced in organizing the focus groups were offset by the advantages of the face-to-face discussion with the target population. For ALSWH, 2 anticipated benefits to utilizing the focus group method were sustained, in regard to testing both the research questions and the Web-based survey. The discussions enabled an exploration of how to engage young women in a longitudinal survey using the knowledge of young women themselves, providing researchers with a nuanced understanding of how to move forward with recruitment. The method also facilitated hands-on testing of the Web-based survey in a neutral setting: a valuable exercise, which identified key areas for survey improvement and simplification. Many focus group participants mentioned that because they had participated in the focus group, they could appreciate the

importance of the study and this would motivate them to stay involved.

In practical terms for health research more broadly, if a research team can convey the value to potential participants of their involvement in the study as part of the recruitment strategy, greater numbers of respondents may be achieved. Further, effective information sharing about the health study can prove useful as part of an overall recruitment strategy. Participants can feel that knowing more about the purpose and processes of the research help them develop a sense of ownership. Survey design features can also assist in data collection. Clever design ideas could offset the perceived effort of completing the survey, such as bright colors and pop-up information boxes. The most common reason for not being able to take part can be that women perceive they have "no time" to help out, and in keeping with focus group findings by Herbert [2] participants prefer a shorter survey to be completed in 1 sitting.

Strengths and Limitations

The paper provides important insight into potential strategies to overcome the difficulty in engaging young women in health research. In terms of lessons learned regarding use of focus groups to inform large-scale health surveys, the current study acknowledges that it is important to reflect upon the recruitment, number, and interactions between participants, which influence the information available to analyze. It is stated that the interaction within groups can generate a particular type of data [13]. The groups conducted for this study varied in size between 1 participant (1 focus group only) and 8 participants. The majority of focus groups were conducted with 2 participants. The variation in participant numbers can mean that a group dynamic wasn't possible between peers of similar age, and that the discussion may have resembled an interview situation rather than a more freely flowing conversation. Important within the broader context of health research is that smaller numbers in focus groups could mean greater assimilation toward a shared view of the matter discussed, as well as with the researchers, leading to lower levels of critical debate about study protocols. Moreover, the current study did not critically appraise how the women spoke, only what they spoke about, meaning that focus group participants' emotions and body language was not factored into the findings.

Although researchers in the current study promoted the focus groups through a variety of outlets, nearly half the participants self-reported that they were currently studying toward, or had completed, a university degree. This could be a reflection of the women's impressions of the importance of the research (ie, whether seen via a hair dressing salon versus a TAFE facility) and whether they felt they were "qualified" to assist. Patton [45] however states that groups can be homogenous in terms of their general characteristics, but this does not necessarily mean they will hold the same attitudes. Snowball sampling used by the current researchers to boost focus group numbers could also mean that participants had prior established relationships, whereas groups are said to work better when participants are strangers [45].

Conclusions

Recruiting young women into health research is challenging. Focus group discussions can help to equip health researchers with targeted interactive access to potential participants' own language and understanding of what health means to them. Our findings point to a strong connection between young people and the Internet, particularly as a mode of communication, and support the move toward large-scale surveys, particularly longitudinal and health-focused surveys, becoming Web-based. Our results provide convincing evidence for the value of asking advice from members of a target population before designing a recruitment strategy, and certainly before commencing recruiting.

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Conflicts of Interest

None declared.

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Abbreviations

ALSWH: Australian Longitudinal Study on Women’s Health
NSW: New South Wales
Qld: Queensland
SMS: short message service
TAFE: technical and further education

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