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Original Paper

The Feasibility, Acceptability, and Efficacy of Delivering Internet-Based Self-Help and Guided Self-Help Interventions for Generalized Anxiety Disorder to Indian University Students: Design of a Randomized Controlled Trial

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Abstract

Background: Generalized anxiety disorder (GAD) is one of the most common mental disorders among university students; however, many students go untreated due to treatment costs, stigma concerns, and limited access to trained mental health professionals. These barriers are heightened in universities in India, where there are scant mental health care services and severe stigma surrounding help seeking.

Objective: To evaluate the feasibility, acceptability, and efficacy of Internet-based, or "online," cognitive behavioral therapy (CBT)-based unguided and guided self-help interventions (using the programs GAD Online and Lantern, respectively) to reduce GAD symptoms in students with clinical and subthreshold GAD and, ultimately, reduce the prevalence and incidence of GAD among the student population.

Methods: Students will be recruited via 3 colleges in Hyderabad, India, and referred for a campus-wide online screening. Self-report data will be collected entirely online. A total of 300 qualifying students will be randomized in a 1:1:1 ratio to receive GAD Online, Lantern, or to be in a wait-list control condition, stratified by clinical and subthreshold GAD symptomatology. Students will complete a postintervention assessment after 3 months and a follow-up assessment 6 months later, at which point students in the wait-list control condition will receive one of the programs. The primary outcome is GAD symptom severity at 3 months postintervention. Secondary outcomes include GAD caseness at 9 months, other anxiety and depression symptoms, self-efficacy, and functional measures (eg, sleep, social functioning) at 3 and 9 months, respectively. Primary analyses will be

differences between each of the intervention groups and the wait-list control group, analyzed on an intention-to-treat (ITT) basis using mixed-design ANOVA.

Results: The study commenced in February 2015. The sample was recruited over a 3-week period at each college. The trial is expected to end in December 2015.

Conclusions: This trial will be the first to evaluate the use of Internet-based CBT programs compared with a wait-list control group for the treatment of GAD among students in Indian universities. If effective, these programs have the potential to reduce the mental health care treatment gap by providing readily accessible, private, and cost-effective evidence-based care to students with GAD who do not currently receive the treatment they need.

Trial Registration: ClinicalTrials.gov NCT02410265 http://clinicaltrials.gov/ct2/show/NCT02410265 (Archived by WebCite at http://www.webcitation.org/6ddqH6Rbt).

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KEYWORDS

Internet-delivered cognitive behavioral therapy (iCBT), generalized anxiety disorder; college health screening; low- and middle-income countries; randomized controlled trial; guided self-help; quality of life

Introduction

Background

Anxiety disorders are the most prevalent class of mental health disorders across the world, with an estimated lifetime prevalence of 4.8-31.0% [1]. Generalized anxiety disorder (GAD), in particular, is one of the most common disorders. In a nationally representative sample in the United States, there was a 5.7% prevalence of GAD [2], and in 14,175 students across 26 United States college campuses, there was a 7% prevalence of GAD [3]. Comparably, in India, a meta-analysis of 13 psychiatric epidemiological studies (N=33,572) conducted in urban and rural India across all age groups yielded an estimated prevalence rate of 5.8% for GAD [4]. Similarly, Nair et al [5] found a 6.6% prevalence of GAD among adolescents in India. In Indian universities, in particular, Sahoo and Khess [6] found a 19% prevalence of GAD in 405 young male university students. These studies suggest that anxiety is a major public health concern, particularly in Indian students. Given that the global average age of onset of GAD is in adolescence and in early adulthood [1,5,7,8], university students are a vulnerable population.

If left untreated, GAD has been demonstrated to have a chronic course and persistent symptoms [9] and is associated with significant distress, disability, quality of life, and medical problems [10]. In addition, subthreshold GAD cases (ie, individuals with significant symptoms who do not meet the full diagnostic criteria for GAD) have also proven equally costly in terms of functional impairment, medically unexplained symptoms such as pain [11], quality of life [12], disability, and help seeking [13,14]. The presence of GAD symptoms even increases the cost of health care from twofold to greater than fourfold [15], with disorder severity positively correlated with total medical costs [16]. Furthermore, both GAD and subthreshold GAD are significant predictors of first onset of other anxiety, mood, substance-use, and impulse-control disorders [17,18]. Studies that have evaluated possible GAD risk factors suggest that subthreshold symptoms might predict onset of GAD [18-20]. Thus, reducing GAD symptomatology among young people and targeting both treatment and prevention have tremendous public-health significance, not least of which is the potential to mitigate ongoing disability and costs [21].

Unfortunately, the majority of affected young people do not receive treatment. Young et al [22] estimated that only 20% of young people in the United States receive adequate treatment. In university environments, students often do not seek treatment due to barriers such as time, stigma concerns, treatment cost, insufficient information about their disorder or available treatment [23,24], and particularly in India, confidentiality concerns [25]. Hunt and Eisenberg [23] discovered that students indicate a preference for self-management and the perception that issues are not serious enough to warrant treatment, and both these are considered primary reasons for not seeking help.

Such stigma around mental health may be a larger issue in Indian versus US populations. Among Asian cultures, research has documented a belief that “emotional reactions” do not merit professional intervention [26]. Another study among Indian adolescents documented a perception that having a mental illness is shameful [27]. Stigma may result in resistance to seeking treatment and those who might seek treatment may be reluctant for fear of discrimination. Online programs offer the advantage of reducing some aspects of stigma.

Even more impactful than the issue of low help seeking is a lack of treatment availability. In the United States, inadequate counselor availability is a significant issue [23]; in developing countries, the issue is significantly worse. In India, for example, there are only 5000 licensed mental health professionals; in the United States, there are 550,000 to treat a population one-fourth of India’s size [28]. This equates to roughly 1 professional per 580 individuals in the United States and 1 per 250,000 in India. There is, therefore, considerable opportunity to improve mental health treatment for students in India by increasing access to treatments that do not require mental health professionals to deliver them.

Internet-based, or “online,” self-help interventions have the potential to overcome barriers such as stigma, cost, and limited specialist services. Online, unguided and guided self-help interventions offer an opportunity to provide cost-effective,

evidence-based care to a large population simultaneously [29]. Unguided, purely self-help (SH) interventions have proven efficacious for treating individuals diagnosed with anxiety disorders including GAD [30-36]. In one study, individuals identified as having clinical GAD received a fully automated SH program and achieved significant improvement across primary symptom severity measures as well as secondary measures such as self-confidence in managing mental health issues and quality of life [32]. Guided self-help (GSH) interventions, in which an online program guide or "coach" [37] supports and guides a user by monitoring progress in the program and providing personalized feedback and encouragement typically via messaging and/or phone, have proven even more effective [38,39]. In fact, GSH interventions have been demonstrated to be as effective as in-person therapy for treating clinical anxiety disorders [39] and depression [40]. These findings suggest that less costly, Internet-based SH and GSH interventions can be considered adequate alternatives to traditional in-person therapy. However, there is no research examining the efficacy of these interventions to reduce GAD symptomatology in students in Indian universities who currently have limited access to mental health care. Therefore, this study seeks to evaluate the feasibility, acceptability, and efficacy of Internet-based interventions, both unguided (using the GAD Online program) and guided (using the Lantern program), to reduce GAD symptoms in Indian university students.

Objectives and Hypotheses

The objective of this trial is to evaluate the efficacy of the GAD Online and Lantern programs for Indian university students with clinical or subthreshold GAD. The primary hypothesis is that the use of either of the active intervention groups, the unguided GAD Online program or the guided Lantern program, for 3 months will lead to greater GAD symptom reduction than will be seen among students in the wait-list control group. The secondary hypothesis is that a guided intervention (Lantern) will lead to greater user engagement and symptom reduction than the unguided intervention (GAD Online). In addition, it is hypothesized that the use of either program will lead to greater functional gains (eg, social functioning) and improvements in mental health self-efficacy than will be seen among students in the wait-list control condition. Table 1 provides a summary of the trial outcome measures.

Methods

Stakeholder Engagement

To better understand the cultural context, existing mental health care system, and needs, a community-based participatory research (CBPR) methodology was used [41]. CBPR calls for a collaborative approach to research that involves all stakeholders in the research process to ensure findings and knowledge gained are meaningful to the community and recommendations are feasible to implement and sustainable. A series of interviews and co-learning discussions were conducted with the following key stakeholders: university administration, existing on-campus counselors and medical professionals, faculty mentors, hostel wardens, and student groups. In interviews with university administration, key topics addressed included current student welfare priorities and where mental health care falls on the list, successful and failed student outreach and engagement efforts, perspectives and concerns of parents, and potential budget for implementing and sustaining new mental health care resources. On-campus counselors provided insight into the typical structure of counseling in Indian school and university environments and the relationship between administration and provider, particularly highlighting that ethics around patient/student privacy are not as widely or consistently practiced in India. Faculty provided the perspective of a "mentor" who primarily gets assigned students flagged for poor academic performance. Hostel wardens provided insight into how "trouble cases" are identified and triaged. Finally, students shared information on student knowledge of resources, barriers that exist to accessing them, and the general campus culture around mental health, particularly noting the stigma around anything involving the term "mental." The details and results of these qualitative assessments will be written up separately.

Preliminary Feasibility and Acceptability Evaluation

To evaluate the feasibility of disseminating these programs to university populations in India, interviews were conducted with university administration to assess the ability to conduct outreach initiatives to engage and educate students about these programs and circulate a campus-wide online survey to assess students' mental health and connect those with significant symptoms to online interventions. Administrators were interested in the proposed survey-linked-to-intervention approach, specifically noting students' interest in opportunities that might allow them to access help privately and on their own time; however, they expressed hesitation about whether students would actually use online programs for mental health care.

To evaluate the acceptability of these Internet-based mental health care interventions among the Indian university student population, a presentation and survey was conducted in April 2014 at one Indian university. Following a presentation to an auditorium of nearly 300 students about anxiety and online programs designed to reduce associated symptoms, students were invited to the adjacent computer laboratory to complete an online survey assessing anxiety symptom severity and student interest in using these types of online programs. The survey was completed by 78 undergraduate students (aged over 18; 52% male). Of these, 94% (73/78) indicated they would consider using "a mobile phone app-based "coached" program" to help them with anxiety symptoms (the Lantern Internet program is mobile optimized so users accessing the website via their mobile phones can have a similar experience to those able to use the iPhone iOS mobile app; therefore, the language "app based" was used). The majority (55/78, 70%) indicated they would like to be contacted should these programs become available at their college.

Of the students who completed the survey, 10% (8/78) had clinical GAD as measured by the 5th Edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5) criterion-based scoring of the GAD-Q-IV [42], and an additional 22% (17/78) had subthreshold GAD, defined as a score of 5.7 or above on the GAD-Q-IV but not being clinical. Additionally, 24% (19/78) had clinical social anxiety as measured by DSM-V

criterion-based scoring of the Social Phobia Diagnostic Questionnaire (SPDQ) [43], 13% (10/78) scored above a clinical cutoff of 8.75 on the Panic Disorder Self Report (PDSR) [44], and 8% (6/78) scored above a clinical cutoff of 38 on the PTSD Checklist for DSM-V (PCL-5) [45]. The prevalence is roughly comparable with that reported by Raakhee and Aparna [46] in a population (N=100) of higher secondary students in India: 13% GAD, 15.6% social anxiety, 15% panic disorder, with 56.8% of all students experiencing 1 or more types of anxiety disorders. Similarly, the prevalence of GAD is comparable with the 19% found by Sahoo and Khess [6] via clinical interviews in a population (N=405) of young adult males in an Indian university. This informal assessment bolstered the belief that the Indian university student population is interested in and might benefit from access to Internet-based interventions addressing anxiety.

Setting

The trial will be conducted in 3 private colleges in Hyderabad, Telangana, a city in the south of India.

Design

A parallel arm, 3 (condition) × 3 (time) randomized controlled trial with equal allocation of students between arms will be used. Students will be randomized to the unguided GAD Online intervention, the guided Lantern intervention, or a wait-list control group. Those assigned to the GAD Online or Lantern group will have full program access for 3 months. Outcomes will be assessed at screening/baseline, postintervention (3 months), and 6-month follow-up (9 months).

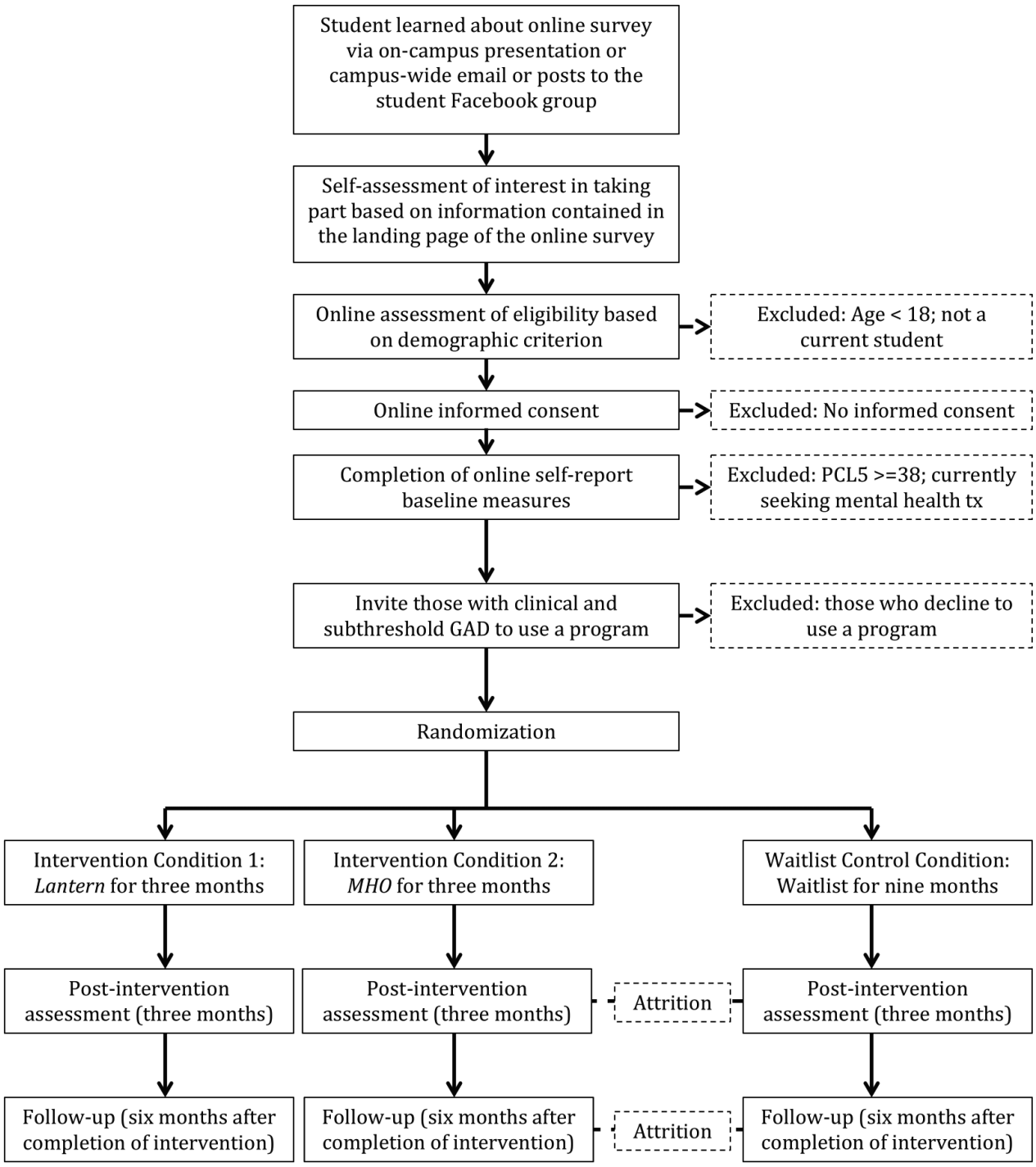
Participants and Procedures

Recruitment

The flowchart (Figure 1) shows the process of recruitment and follow-up of students in the trial. Potential participants will be recruited through an online survey delivered across 3 colleges in Hyderabad. The research coordinator will visit each site and deliver presentations about the study as well as send campus-wide emails. After raising awareness about the study across the campus, the research coordinator will circulate a link to an online survey via email, which will also be posted in the student Facebook page and other social media groups. The landing page will again provide information about the study and instruct interested individuals to move forward to complete an online consent form and screening survey. This landing page will also contain the contact information of the on-campus counselor, as this information is meant to be available to all students across campus.

Those who meet criteria for clinical or subthreshold GAD will be invited to participate in the study and use an online program. Those who are interested will be informed that they will be assigned to a condition within 4 weeks. Everyone will receive their assignment email within 4 weeks following baseline, and those assigned into the 2 treatment conditions may activate their program account and begin immediately. Students in the GAD Online or Lantern conditions will have access to the intervention over a 3-month period. Students assigned to GAD Online can access content at whatever pace they choose. Students assigned to Lantern can access up to1 session per day because the program is structured to build mastery. Program access will be disconnected following this period. If students still meet clinical criteria for GAD at the end of the intervention, they will receive a referral to visit the on-campus counselor. Students will also be given a list of free, publicly available online self-help resources for anxiety (eg, Mental Health Online, ThisWayUp). Continuation in the guided intervention will not be possible due to resource constraints around program coach availability in the context of this research trial. Assessments will be conducted completely online. At each assessment time point, students will receive an email with a link to an online survey hosted on Qualtrics, an online survey platform with industry-standard data security measures.

Figure 1. Trial flow chart.



Eligibility

Eligible participants will be current students of the colleges, all of which are English medium, aged 18 or older who provide their email address, informed consent to complete the study, and meet criteria for clinical or subthreshold GAD. This age range was chosen because individuals younger than 18 require parental consent for participation in research. Based on self-report responses in the online survey, students are categorized into clinical, subthreshold, and asymptomatic for GAD. Those who meet DSM-V diagnostic criteria for GAD as measured by criterion-based scoring of the 4th edition of the Generalized Anxiety Disorder Questionnaire (GAD-Q-IV) [42] are classified as clinical, or more realistically,

"a probable diagnosis of GAD based on self-report." To note, the diagnostic criteria did not change from DSM-IV to DSM-V, and therefore the GAD-Q-IV questions can still be used to assess DSM-V GAD criteria. Those who score 5.7 or above using dimensional scoring of the GAD-Q-IV but are not clinical are classified as "subthreshold." All others are asymptomatic. It is also important to note that clinical diagnosis should be established using a clinical interview, which is not feasible in this study, thus the use of the self-report measures. Moreover,

this study's aim is not to provide diagnoses to students but rather to use gold-standard instruments to provide appropriate "matches" between students and available resources (ie, the research study or referral).

The PTSD Checklist for DSM-V (PCL-5) [45] will be used to refer individuals who self-report clinical symptoms of PTSD to targeted intervention or clinical services. CBT for GAD has been demonstrated to improve symptoms across comorbid disorders such as social anxiety disorder and depression [47], and therefore a "pure GAD" sample is not necessary. However, PTSD requires a more specific approach, and data on CBT for GAD's impact on PTSD are not available. A more conservative approach was followed: PTSD "probability" based on the PCL-5 (ie, scoring 38 or above) will result in a referral to seek help from the on-campus counselor and to access the free, publicly available iPhone and Android mobile app, PTSD Coach [48]. It is difficult to get a "clean sample," so a balance between safety and generalizability was pursued. No additional symptom-based exclusion criteria were imposed, because a primary goal of this study was to connect most students to a service they might be willing to access. Once a student is connected to a program, mechanisms exist to ensure referrals are given if symptoms are not adequately reduced and/or other symptoms present themselves (eg, program coaches give referrals to those in the guided Lantern intervention and postintervention survey feedback gives referrals to those who still report clinical symptoms). Any individuals who report currently receiving mental health treatment will also be excluded from enrolling in the study and receiving a program. Following rule-out, individuals with a clinical or subthreshold GAD classification will be invited to participate in the study evaluating online interventions. Those who accept will be randomized to one of three conditions: a self-help program (GAD Online), a guided self-help program (Lantern), and a wait-list control group. A complete list of inclusion/exclusion criteria can be found in Textbox 1.

Textbox 1. Inclusion and exclusion criteria for the study.

Inclusion criteria

- 18+ years old
- Current student at the university
- Meet DSM-V criteria using criterion-based scoring on the GAD-Q-V (clinical) OR score ≥ 5.7 using dimensional scoring of the GAD-Q-IV but not meet DSM-V clinical criteria (subthreshold)
- Provide an email address
- Consent to participate in the study

Exclusion criteria

- Currently receiving mental health treatment
- Current diagnosis of PTSD (PCL-5 score ≥ 38)

Ethical Concerns and Consent

The trial protocol has been granted ethical approval by the Institutional Review Board at Stanford School of Medicine (protocol number 31629) and governing bodies at each of the participating colleges. The protocol is registered with ClinicalTrials.gov (NCT02410265).

In the online consent form, students are notified of how their privacy and confidentiality of their data will be maintained. All survey data will be gathered and stored on Qualtrics, an online survey platform with industry-standard data security measures. Students are informed that their participation in the survey or in a program is private and will not be shared with their college, their parents, their peers, etc. Furthermore, all data gathered will be aggregated and deidentified prior to use in any publications. Only the research coordinator has access to identifying student data. Program coaches assigned to work with students in the guided Lantern intervention connect with students via the secure program platform; no personal contact information (eg, email, phone number) is exchanged. As Lantern users receive email notifications when they receive a message from a program coach, Lantern has access to the student users' emails; however, Lantern is HIPAA compliant and follows appropriate data management and sharing protocols. Students assigned to the GAD Online intervention are provided with generic account user names and passwords, meaning GAD Online never has need for or access to students' email addresses.

Baseline Assessments

After giving online consent, students complete the baseline assessment, which also serves as the screen for eligibility. Table 1 provides a summary of the measures that will be used.

Randomization

Randomization will be carried out by the research coordinator after the baseline assessment. Random allocation to the treatment groups will occur within 4 weeks after the screening/baseline survey has been completed. The algorithm for random allocation will consist of a stratified block design, with stratification by level of symptoms (clinical or subthreshold) and a block size of 6. There will be 2 strata, corresponding to clinical/subthreshold symptom level. Allocation will be administered using Randomizer, a Web-based patient randomization service for clinical trials.

Risk Management Protocol

Risk management procedures were developed collaboratively with the participating universities, on-campus counselors, online program providers (ie, GAD Online and Lantern), and the research team. All students will be provided with the contact information for the respective on-campus counselor at the start and finish of the online survey. In this way, anyone exposed to the survey will be informed about currently accessible services. Furthermore, all universities have agreed to increase the current time of the existing on-campus counselor should student demand require it. In addition, for any individual in the guided intervention (Lantern), program coaches will monitor for worsening symptoms and express student preference to receive more intensive treatment and provide a referral to the on-campus counselor as necessary. Additionally, if a student in the guided program indicates thoughts of harm to self or others, the coach will notify the coach clinical supervisor (a clinical instructor at Stanford and licensed mental health care professional in the United States) via email and the campus-specific, on-site counselor via phone. Both will help determine how best to manage the situation, and the on-campus counselor will intervene and reach out to the student as necessary. In the consent, students are informed that confidentiality can be breached should the student indicate harm to self or others. Those in the unguided intervention will be explicitly informed that their actions in the program are not monitored and that they should contact the on-campus counselor should they want live support. They will again be reminded of the contact information of the on-campus counselor when they are assigned into this unguided intervention.

Sample Size Estimation

The sample size estimation is based on detecting differences in GAD symptom severity change (measured using dimensional scoring of the GAD-Q-IV) from baseline to post- and follow-up assessments between each active conditions and the wait-list control group. Treatment trials of online, CBT-based, guided self-help interventions for GAD have found an average large effect size of roughly 1.07 relative to a wait-list control group using the GAD-Q-IV [38]. In this study, more than half of the treated participants had recovered according to a structured interview, suggesting that a treatment effect size of 1.07 is clinically meaningful. Treatment trials of purely self-help interventions for subthreshold and clinical anxiety have found smaller yet still promising effect sizes ranging from 0.62 to 0.84, respectively, relative to a wait-list control group [31].

To enable detection of difference in mean change between each active intervention and the control group, the sample size is powered to detect at least a medium effect size of 0.5. With 80% power and assuming a 5% significance level, a minimum total sample size per group of 64 is required. Assuming the GAD-Q-IV SD is 3.46 [49], the proposed sample size provides 80% power to detect a between-group effect corresponding to a GAD-Q-IV change of 1.73 points (or a Cohen's *d* of 0.5 or greater). By targeting 100 students recruited per condition (N=300), even allowing for nearly 30% attrition, which is a possibility in Internet-based intervention trials, GAD-Q-IV mean differences between each active condition compared with the wait-list control condition will be detected with sufficient power. The difference in mean change between the two active conditions is not a primary comparison, as they are two different programs.

The Interventions

Active Condition 1: "Lantern" Guided Self-Help Intervention

The Lantern Anxiety Program is a cognitive-behavioral intervention (CBT) that can be accessed via any Internet-enabled computer, mobile phone, or tablet via the Go Lantern website (Figure 2). The program includes psychoeducational content, interactive tools and exercises, symptom monitoring, and an online program coach who can monitor a user's progress in the program and provide personalized feedback and encouragement via in-program messaging and voice calls. The program is based on an evidence-based, 14-session CBT for GAD intervention, developed by Dr Michelle Newman [36,50,51]. CBT for GAD has been demonstrated to produce the largest effect sizes when compared to other therapy conditions such as analytic psychotherapy and nondirective therapy [52]. The CBT for GAD treatment includes (1) applied relaxation training, which involves the identification of early cues of anxiety, learning the skills of progressive relaxation (PR) and other relaxation techniques and learning how to apply relaxation to anxiety cues (called "applied relaxation," [AR]); (2) imaginal rehearsal via self-control coping desensitization, using AR and alternative, nonanxious coping thoughts; and (3) cognitive therapy methods to help change how clients perceive, interpret, and believe, so that they will see less threat in the world and feel more confidence in their abilities to cope with the future.

Content is divided into 8 units, each with 5 sessions, resulting in 40 daily 10-minute sessions. The 8 units comprise an introduction to anxiety, automatic thoughts, cognitive reframing, introduction to behavior change, imaginal exposure, situational exposure, mindfulness, and habit formation. There are both cognitive (eg, worry tool, mindfulness) and behavioral (eg, progressive muscle relaxation, deep breathing, guided imagery) techniques taught in each module and always accessible thereafter. Users can access one session per day, with the next session unlocking only after the prior session is completed; therefore, less engaged users may not be exposed to all content. The program and program coach prompt users to schedule reminders to facilitate self-monitoring, encourage completion of scheduled activities, and promote use of tools and techniques in the program. Users can schedule reminders to complete sessions and to use techniques or self-monitoring entries at their desired frequency. They are prompted to do so after learning a new technique. Coaches can also schedule reminders for themselves to send users messages.

Students using Lantern will be guided through the program by mental health workers in India who have been trained to serve as online program coaches. To be eligible to be a coach, individuals must have some background and/or experience in psychology, have at least 6 months of supervised, in-person counseling experience, own a personal computer or smart device and check email daily, be comfortable with technology, and

have a minimum of 3 hours per week to dedicate to coaching. A total of 20 potential program coaches were recruited, all of whom had completed a 6-month training course in the basic principles of psychological counseling from the Hyderabad Academy of Psychology based in Hyderabad, India.

A pretraining assessment was used to assess existing skill levels and inform training content development. To begin, 4 weeks of remote training covering basic foundational concepts was conducted. Manuals and papers reviewing CBT for GAD, panic disorder, and social anxiety were assigned, and a virtual discussion session was conducted each week via online collaboration platform GoToMeeting. In addition, coaches were provided a demo user account of the Lantern Anxiety Program so that they could familiarize themselves with the intervention content. Following remote training primarily reviewing content and basic concepts, a 3-day in-person training was conducted in Hyderabad, India, in December 2014. During this training, the Lantern coaching platform was introduced and coaches learned how to effectively interact with the coaching dashboard. The coaching dashboard enables coaches to monitor the activity and engagement of the students assigned to them as well as to send messages to them. Coaches were instructed on guided self-help coaching best practices, introductory phone call and messaging protocols, motivational interviewing, and risk management procedures.

Following training, coaches were assessed for comfort with technology via direct observation of their interaction with the Lantern coaching dashboard during interactive role plays. Coaches who demonstrated low technology fluency and experienced usability issues interacting with the coaching software in comparison with the group were asked to attend additional training and practice sessions to continue. Behavioral rehearsal tasks were used to evaluate coaches' fidelity to the coaching protocol. Coaches were required to complete a mock introductory phone call with a control student (played by the research coordinator or volunteer students following a script), and fidelity to the phone call procedural checklist was assessed. Coaches were also required to respond to message prompts designed to assess their mastery of core competencies of CBT for GAD in the context of a guided self-help intervention. Their responses were rated against a messaging best practices checklist that was developed by the authors. Finally, coaches had to complete an established "e-therapist assessment" developed by the National eTherapy Centre (NeTC) at Swinburne University of Technology [53] and score at least 80%. Of the 20 trainees who began training, 65% (n=13) cleared the post-training evaluation and moved forward to become program coaches for this trial.

Throughout the intervention period, coaches will have on-going monitoring and support. Coaches must attend a weekly, live 1-hour supervision, during which coaches can present any challenging cases to the group and receive feedback from the coach clinical supervisor (a clinical instructor at Stanford and a licensed mental health care professional in the United States). Coaches will also be asked to submit 1 challenging message exchange per week to a clinical psychology doctoral student for individual feedback on messaging style and content. Finally, coaches can post questions and messages on a group listserv to get feedback from the supervisor as well as from peers.

Active Condition 2: "GAD Online" Pure Self-Help Intervention

The GAD Online program is also CBT based and can be accessed via any Internet-enabled computer, mobile phone, or tablet via the Mental Health Online website (Figure 3). Mental Health Online is a suite of online mental health programs developed and maintained by the NeTC at Swinburne University of Technology and funded by the Australian Federal Government Department of Health. The GAD Online program delivered in this trial, which was previously reported on as an Anxiety Online module [33], provides psychoeducation about GAD and techniques to reduce GAD symptoms (eg, relaxation, challenging thoughts, coping with worry, problem solving). The content covered in this program is comparable to that delivered in the Lantern GSH program. Content is divided into 12 modules, with 1 module per week being the suggested use; however, users can progress at their own pace. Unlike the design of the Lantern program, users can access other modules in the GAD Online program even if they do not complete the first module. Students are explicitly told they are not monitored and should contact the on-campus counselor should they require real-time support from a professional.

Figure 2. Screenshot of Lantern Internet-based user view.

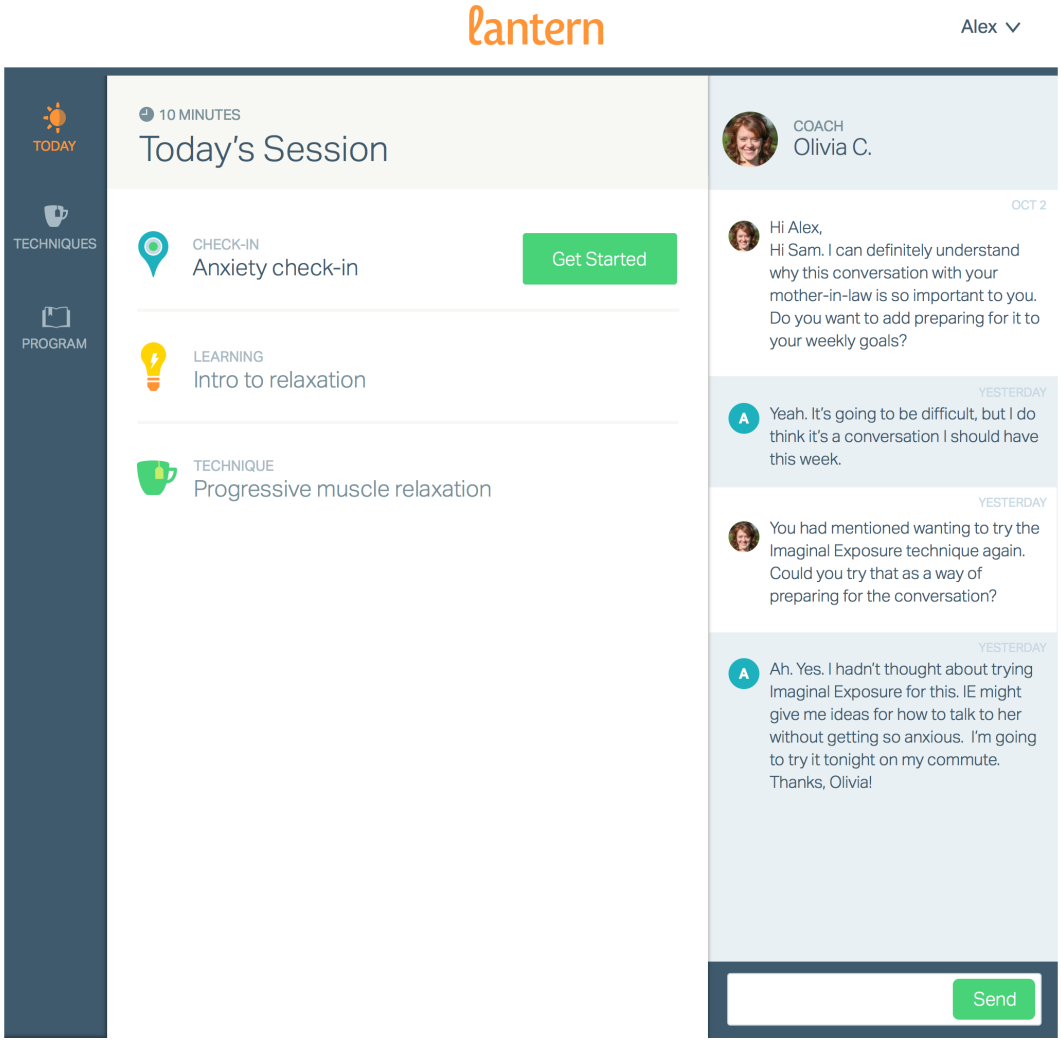
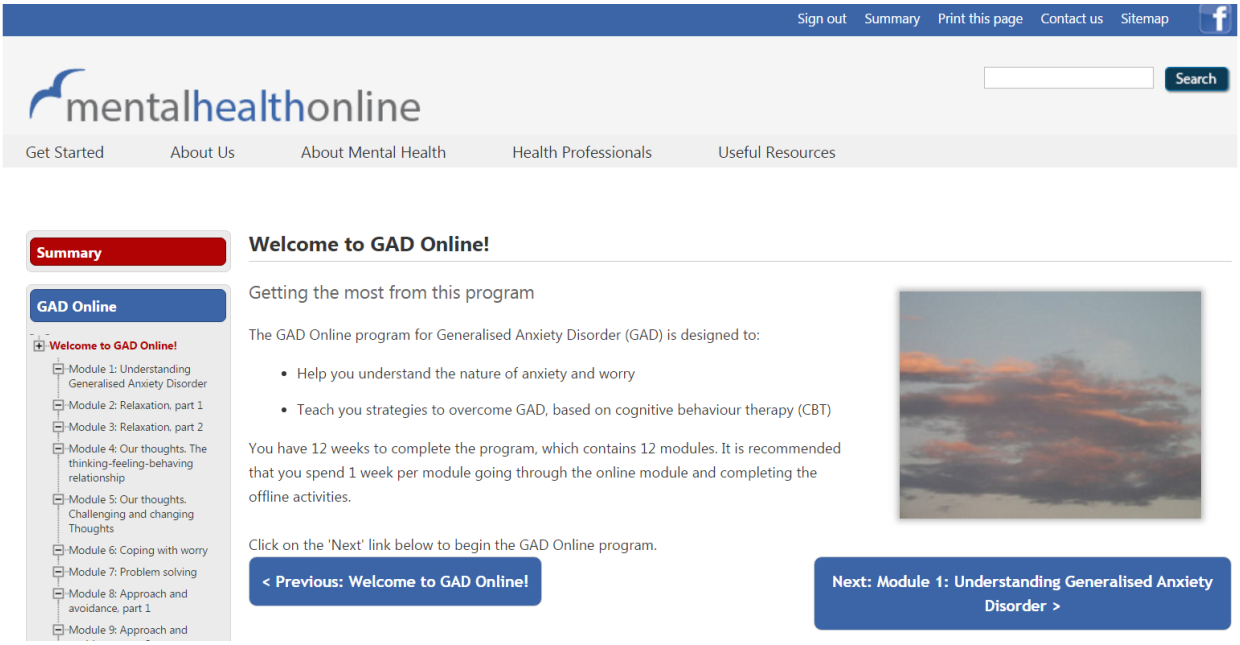


Figure 3. Screenshot of Mental Health Online's GAD Online program.



Wait-list Control Condition: Delayed Program

Students randomized to the wait-list control condition will complete surveys at the postintervention time point (3 months) and the follow-up time point (9 months). Following successful completion of these surveys, they will be allocated to one of the online interventions. Students will be assessed for whether they sought help from the on-campus counselor at each time point. The counselor's contact information is available to all students across the campus.

Minimization of Contamination

As randomization occurs within each college campus, there is a possibility of contamination, in that students could share their programs with each other. However, this type of contamination is not believed to be likely, as the programs are presented as personalized and private. Furthermore, because recruitment for this exploratory trial is occurring at 3 distinct colleges, selected more so due to partnership, comparability between sites is not known. Future, larger trials might consider randomization by college site.

Measures

Outcome data will be collected at post-treatment (3 months) and 6-months follow-up (9 months). The 3-month outcome is the primary end point as the intervention delivery will be completed and the optimal effect of the treatment would be expected. The 6-month follow-up is included to evaluate the sustainability of the effect of the intervention. The outcome assessment measures are summarized in Table 1. Although the primary outcome measure is the GAD-Q-IV, we also assessed other anxiety disorders and potential functional impairments. Because only a few studies have gathered comprehensive data on prevalence rates of types of anxiety disorders and given the unique opportunity to administer an online survey to this large population, a subaim of this study and the involved stakeholders is to gather epidemiological data that can inform future research and policy and programmatic decisions by stakeholders to address the needs of students. Besides, given the high degree of comorbidity present among individuals who experience GAD [6,36,54], measuring psychological state and evaluating co-occurring disorders such as other anxiety disorders and depression are important. Finally, because CBT for GAD has been demonstrated to address symptoms associated with other disorders [47,55], both the prevalence and secondary outcomes are relevant. To ease anticipated survey burden, the survey is designed to use logic that allows for participants with low scores and fewer comorbid concerns to "skip" subsections. Demographic data will include gender, age, race/ethnicity, sexual orientation, religion, relationship status, family income, and hometown.

Details of the Measurements

Generalized Anxiety Disorder Questionnaire (4th Edition)

The primary outcome is GAD symptom severity as measured by dimensional scoring of the 4th edition of the Generalized Anxiety Disorder Questionnaire (GAD-Q-IV) [42]. GAD caseness, as measured by categorical scoring of the GAD-Q-IV, is a secondary measure. The GAD-Q-IV is a 9-item self-report measure designed as an initial screen for the presence of GAD based on the DSM-IV. The GAD-Q-IV showed 89% specificity and 83% sensitivity when compared to structured interview diagnoses of individuals with GAD, social phobia, panic disorder, and a nonanxious comparison group. The GAD-Q-IV has demonstrated good test-retest reliability in a college sample over a 2-week assessment, with 92% of the sample showing stability across time with respect to GAD diagnosis [42].

Emotional Distress From Anxiety Measure

The Patient Reported Outcomes Measure Information System (PROMIS) Emotional Distress from Anxiety measure [56] is a reliable 8-item self-report measure to evaluate emotional distress from anxiety. This measure was included as a potential global outcome measure of improved quality of life.

Penn State Worry Questionnaire

The Penn State Worry Questionnaire (PSWQ) [57] is a 16-item self-report measure of the frequency and intensity of worry. The PSWQ has been shown to distinguish individuals with GAD from individuals with other anxiety disorders [58]. This measure was included to assess change in worry, a primary symptom of GAD.

Panic Disorder Self-Report

The Panic Disorder Self Report (PDSR) [44] is a 22-item self-report measure designed to diagnose panic disorder based on DSM-IV criteria. The PDSR showed 100% specificity and 89% sensitivity when compared with clinician-based ADIS-IV-L diagnoses of individuals diagnosed with panic disorder, GAD, social phobia, and a nonanxious comparison group. This measure, along with the other measures of anxiety disorders and depression, was included to assess both prevalence of this type of anxiety and the impact of the active interventions on symptoms associated with typically co-occurring disorders.

Social Phobia Diagnostic Questionnaire

The Social Phobia Diagnostic Questionnaire (SPDQ) [43] is a 10-item self-report measure designed to diagnose social phobia based on DSM-IV criteria. The SPDQ showed a specificity of 82% and a sensitivity of 85% when compared with clinician-based structured interview diagnoses of individuals meeting criteria for social phobia, panic disorder, and a nonanxious comparison group.

Post-Traumatic Stress Disorder Checklist

The Post-Traumatic Stress Disorder Checklist for DSM-V (PCL-5) [45] is a 20-item self-report measure that assesses the 20 DSM-V symptoms of PTSD. It was designed to monitor symptom change during and after treatment, screen individuals for PTSD, and make provisional PTSD diagnoses.

Table 1. Scales to be administered at each time point.

Outcome variable			Measurement tool/data collection method	Time point (months)		
				0	3	9
Eligibility			Self-report	X		
Demographics			Self-report	X		
Field of study			Self-report	X		
Perceived social support			Self-report	X		
Belief in efficacy of online programs (credibility/expectancy)			Self-report	X		
Contamination			Self-report		X	
Grade point average			Self-report	X	X	X
Primary outcome						
	Difference in GAD symptom severity at postintervention and follow-up between the intervention groups and control group		4th edition of the Generalized Anxiety Disorder Questionnaire (GAD-Q-IV) (scored dimensionally)	X	X	X
Secondary outcomes						
	Anxiety					
		GAD caseness	GAD-Q-IV (scored categorically)	X	X	X
		Emotional distress from anxiety	PROMIS Emotional Distress from Anxiety measure	X	X	X
		Worry	Penn State Worry Questionnaire (PSWQ)	X	X	X
		Panic disorder symptoms	Panic Disorder Self-Report (PDSR)	X	X	X
		Social anxiety symptoms	Social Phobia Diagnostic Questionnaire (SPDQ)	X	X	X
		PTSD symptoms	Post-Traumatic Stress Disorder Checklist for DSM-V (PCL5)	X	X	X
		OCD symptoms	LEVEL 2—Repetitive Thoughts and Behaviors—Adult (adapted from the Florida Obsessive-Compulsive Inventory [FOCI] Severity Scale [Part B])	X	X	X
		Distress severity for specific fears	Self-report	X	X	X
	Depression					
		Depression symptoms	Depression Anxiety Stress Scales (DASS21)	X	X	X
	Behavioral measures					
		Alcohol consumption	Self-report	X	X	X
		Help seeking	Self-report	X	X	X
		Medications	Self-report	X	X	X
	Functional measures					
		Sleep	Insomnia Severity Index (ISI)	X	X	X
		Difficulties with emotions and relationships	Strengths and Difficulties Questionnaire (SDQ)	X	X	X

Outcome variable		Measurement tool/data collection method	Time point (months)		
			0	3	9
	Satisfaction with social roles	PROMIS Satisfaction with Social Roles and Activities measure	X	X	X
	Nonspecific distress	Kessler Distress Measure (K10)	X	X	X
	Confidence in ability to manage anxiety symptoms	General Perceived Self-efficacy Scale (GSE)	X	X	X
	Motivation to work on anxiety	Self-report	X	X	X
Process measures					
	Program use and engagement	Self-report		X	
	Usefulness of GAD Online or Lantern	Self-report		X	
	Satisfaction with GAD Online or Lantern	Client Satisfaction Questionnaire (CSQ)		X	

Repetitive Thoughts and Behaviors

The LEVEL 2—Repetitive Thoughts and Behaviors—Adult (adapted from the Florida Obsessive-Compulsive Inventory [FOCI] Severity Scale [Part B]) [59] assesses obsessive-compulsive disorder symptoms. This emerging measure of the DSM-V was developed to be administered at the initial patient interview and to monitor treatment progress.

Depression Anxiety Stress Scales-Short Form

The Depression Anxiety Stress Scales-Short Form (DASS21) [60] is a 21-item self-report measure designed to measure negative emotional states of depression, anxiety, and tension or stress. The DASS is divided into 3 self-report scales. The depression scale assesses dysphoria, hopelessness, devaluation of life, self-deprecation, lack of interest/involvement, anhedonia, and inertia. The Anxiety Scale assesses autonomic arousal, skeletal muscle effects, situational anxiety, and subjective experience of anxious affect. The Stress Scale is sensitive to levels of chronic nonspecific arousal, assessing difficulty relaxing, nervous arousal, and being easily upset/agitated, irritable/over-reactive, and impatient. All scales have been shown to have high internal consistency and to yield meaningful discriminations in a variety of settings.

Insomnia Severity Index

The Insomnia Severity Index is a 7-item self-report questionnaire to assess insomnia severity [61]. It has adequate internal consistency and is a reliable self-report measure to evaluate perceived sleep difficulties. This measure, along with the other measures of general functioning, was included to assess the impact of the active interventions on functional impairments typically associated with GAD.

Strengths and Difficulties Questionnaire

The Strengths and Difficulties Questionnaire [62] assesses self-reported difficulties with emotions, concentration, and relationships. It is a reliable short-form, self-report assessment of inattention, peer relationships, and pro-social behavior, factors that, if improved, could indicate positive impact of the active interventions.

Satisfaction With Social Roles

The PROMIS Satisfaction with Social Roles and Activities measure [63] is an 8-item self-report assessment of satisfaction with one's ability to perform daily activities and meet the needs of various relationships.

Kessler Distress Measure

The Kessler Distress Measure [64] assesses nonspecific distress. The 10-question (K10) self-report scale has good precision as well as consistent psychometric properties across major sociodemographic subsamples. This measure was included as an assessment of a global outcome of reduced general distress.

General Perceived Self-Efficacy Scale

The General Perceived Self-Efficacy scale (GSE) [65] is a 10-item self-report measure that assesses beliefs in one's capability to handle new and difficult tasks and adaptive challenges after experiencing stressful life events. This measure was included because enhanced self-efficacy is considered one of the main psychological benefits of self-help interventions [66].

Program Use and Engagement

Program usage will be examined for the 2 active intervention groups with respect to 3 indices: frequency of logins, frequency of self-monitoring, and number of modules accessed. Reason

for drop out will be assessed via self-report questions in the survey as well as a semistructured interview with those who are willing to enroll in a follow-up substudy. For both the GAD Online and Lantern platforms, module access can be passively monitored. Engagement in Lantern can be further qualified in terms of engagement with the program coach. The Lantern software can program reminders for coaches to reach out to inactive users after a specific period. Coaches are given training in motivational interviewing and encouraged to use these skills to help engage users and address fluctuating stages of change during the intervention. Unstructured data (ie, text) from users' program entries and correspondence with coaches can be qualitatively (eg, language used) and quantitatively (eg, user: coach message frequency) evaluated, with participant consent and understanding that message content can be used for research, to examine motivational strategies and engagement.

Program Satisfaction

Program satisfaction will be examined for the 2 active intervention groups using the Client Satisfaction Questionnaire (CSQ) [67]. The CSQ is an 8-item self-report statement of satisfaction with health and human services. The generic questionnaire has been customized for each intervention (eg, replacing the term "service" with "the GAD Online program" or "the Lantern Anxiety Program"). Those using Lantern will also provide feedback on their experience working with the program coach.

Data Management

All data will be stored on secure servers hosted by Qualtrics, and data downloaded from those servers will be managed and stored on encrypted computers accessed only by members of the Stanford research team.

Analysis

Descriptive Analyses

Chi-squares (categorical variables) and *t* tests (continuous variables) will be used to compare demographic variables and baseline scores on the outcome measures for the 3 groups. Findings will be reported according to the CONSORT guidelines [68], including a trial flowchart. This will include total students assessed for inclusion and exclusion criteria within period of screening, number of students meeting inclusion or exclusion criteria, number screened for eligibility, number agreeing to enter the intervention trial, and number refusing or excluded (with reasons). The number continuing through the trial, actively withdrawing, and passively lost to follow-up will be shown by arm. The outcome measures will be summarized at baseline/screening, at 3-month and 9-month follow-up by the intervention arm and overall.

Outcome Analyses

A 3 condition × time mixed-design ANOVA will be conducted to examine both primary and secondary hypotheses. Using a mixed-design ANOVA will test for differences between 2 or more independent groups while subjecting participants to repeated measures. The model is a type of mixed effect model, with the fixed effects factor being a between-subjects variable and the random effects factor being a within-subjects variable. Follow-up paired comparisons will be conducted to specifically assess if there is significant differential change in GAD symptoms between each active condition and control from the baseline to the postcondition and follow-up assessments. Within- and between-group effect sizes will be calculated using Cohen's *d* (based on the pooled standard deviation), specifically contrasting each active condition with the control condition for the main outcome.

Primary analyses will be undertaken using an intention-to-treat (ITT) approach, including all participants randomized regardless of treatment actually received, program engagement, or withdrawal from the trial. Using a mixed model design will allow us to include participants with missing data, which is likely to occur in large Internet-based trials.

Study site will be included as a covariate as this might have an influential effect given that the 3 sites were selected primarily based on accessibility versus between-site comparability. If some sites have poor recruitment, the data will be combined.

Mediator and Moderator Analyses

Potential mediators and moderators of the efficacy of the intervention will also be explored. For example, level of program engagement, defined by number of sessions completed and number of days logged on to the program, will be evaluated for mediation of outcomes.

Additionally, given that randomization was stratified between clinical and subthreshold populations, whether symptom severity influences the efficacy of the intervention to reduce GAD symptoms can be explored. Additional predictors of outcome that will be explored include medication use, motivation to work on anxiety, belief in efficacy of online programs, perceived social support, and grade point average. Medication use will be controlled for in the analyses if the sample is large enough; otherwise, the results will be provided qualitatively.

Results

The study commenced in February 2015. The sample was recruited over a 3-week period at each college. The trial is expected to end in December 2015.

Discussion

Potential Impact

This trial represents an opportunity to explore whether technology can be leveraged to reduce the significant mental health treatment gap in India, particularly for students in Indian universities. This will be the first trial to examine the feasibility, acceptability, and efficacy of Internet-based, unguided and guided self-help interventions to reduce GAD symptoms in Indian university students. It will also be the first trial evaluating the acceptability and feasibility of online, guided self-help interventions supported by trained mental health workers in India.

Intervening to reduce GAD symptoms in students is important because increasing GAD symptomatology is associated with greater functional impairment [11], reduced quality of life [12],

greater disability and help seeking [13,14], and greater costs of health care [15]. Furthermore, without treatment, GAD symptoms have a chronic course and persistent symptoms [9]. Given the increasing prevalence of Internet connectivity and mobile phone penetration [69] and in the face of limited mental health care professional availability, delivering evidence-based mental health care via the Internet may be particularly attractive to individuals in developing countries, particularly students in Indian universities, who have limited access to mental health care services. Furthermore, both programs that will be employed, GAD Online and Lantern, are scalable platforms, thus making the use of Internet-based, unguided and guided self-help interventions a practical approach. If the interventions are found to be effective, these programs can be more specifically adapted for this population and then be promoted and disseminated seamlessly to the larger student population, universities, and younger individuals.

Limitations

Relying entirely on self-report data to assess individuals is a limitation of this study and implies that the allocation of interventions may not be as reliable as if diagnostic interviews and other objective measures were used. This method was selected for two reasons. First, one of the primary barriers to help seeking is stigma, particularly in this population. In stakeholder discussions, both administrators and students highlighted that concerns around confidentiality and disclosure to others – peers, professors, parents – prevent many students from visiting the existing on-campus counselor. Thus, a private, entirely online approach is likely to achieve greater participation and more accurate responses. Second, one of the main goals of this model of intervention is to develop a cost-effective and sustainable way to disseminate and evaluate these programs at scale, and requiring an in-person assessment would dramatically increase costs and reduce sustainability. Moreover, these assessments are meant to inform recommendations about program "matches" that meet participants' self-reported needs, and participants are empowered to choose the avenue of support they perceive to be the best fit (ie, on-campus counselor or online program). Following initial trials establishing feasibility, clinical interviews can be conducted along with self-report assessments to evaluate the validity and reliability of the self-report screening measures for this population. Additionally, in future trials, the real and important limitation of response burden could be addressed and fewer, more targeted measures could be administered.

Another limitation is that the primary outcome measurement tool, the GAD-Q-IV, and the associated diagnostic thresholds have been normed in US university student populations but not in an Indian university student population. One implication is that the prevalence of the disorder could be mischaracterized due to diagnostic criteria being interpreted differently cross-culturally. For example, the DSM-IV GAD criterion that anxiety be “excessive” might lead to underdiagnosis of GAD in developing countries in which worry is less likely to be reported excessive in the presence of comparatively more severe life concerns [70]. Using dimensional versus categorical scoring of the GAD-Q-IV to both determine eligibility and monitor for change in symptomatology, this concern might be partially addressed. The cutoff point of 5.7 was found to be overly sensitive in the US college population [42], so it is a reasonable threshold above which to initially allocate interventions to those potentially in need but not currently accessing services. However, future and parallel research should focus on validating the assessment tools.

Another potential limitation is the fact that the two active conditions differ both by the degree of coaching support and the program used, which precludes this study from clearly deducing the additional, if any, benefit of guidance. However, the primary hypothesis is that the use of one or both of the active interventions will lead to a greater GAD symptom reduction than the wait-list control. If the unguided program is found to be both feasible to disseminate to this population and relatively effective, future controlled studies can evaluate the comparative benefit of guidance and conduct cost-benefit analyses of delivering unguided versus guided programs. The conditions under which it is more or less effective will also be explored (eg, it might be more effective for those with higher reported motivation who are more likely to complete more of the program). The primary initial aim is an evaluation of the feasibility and efficacy of the model of survey-linked-to-online-interventions to reduce the incidence and prevalence of common mental health disorders in defined Indian university populations.

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Authors' Contributions

NK, MGN, and CBT contributed to the conceptualization, development, and design of the study as well as the writing of the manuscript. JIR and EK contributed to the conceptualization and development of the study. MM contributed to the design of the study, specifically providing input regarding the treatment of anxiety among youth in India, as well as manuscript revision. MJ contributed to the design of the study, specifically providing input regarding how the Lantern technology could be used. NT and JMA contributed to the design of the study, specifically providing input regarding how the Mental Health Online technology could be used. SS contributed to the design of the study, specifically providing input regarding how student engagement, recruitment, and risk management could be executed in the Indian university context.

Conflicts of Interest

MGN and CBT are on the scientific advisory board of Lantern, which developed the guided self-help anxiety program. However, neither author derives personal financial benefit from the operation of Lantern. MJ is employed at and has equity in Lantern, which is a for-profit company that distributes one of the interventions described in this study. MJ was involved in study design related to using the Lantern technology. She does not have any direct contact with participants. She will not be handling any data nor will she be involved in data analysis. The other authors have no conflicts of interest to declare.

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Abbreviations

CBT: cognitive behavior therapy
DASS: Depression Anxiety Stress Scales
DSM: Diagnostic and Statistical Manual of Mental Disorders
FOCI: Florida Obsessive-Compulsive Inventory
GAD: generalized anxiety disorder
GAD-Q-IV: 4th edition of the Generalized Anxiety Disorder Questionnaire
GSES: General Self-Efficacy Scale
ISI: Insomnia Severity Index
K10: Kessler distress measure
PCL-5: Post-Traumatic Stress Disorder Checklist for DSM-V
PDSR: Panic Disorder Self Report
PROMIS: Patient Reported Outcomes Measure Information System
PSWQ: Penn State Worry Questionnaire
RCT: randomized controlled trial
SDQ: Strengths and Difficulties Questionnaire
SPDQ: Social Phobia Diagnostic Questionnaire

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Original Paper

Optimizing Social Network Support to Families Living With Parental Cancer: Research Protocol for the Cancer-PEPSONE Study

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Abstract

Background: Parental cancer can have a significant impact on a family's psychosocial functioning and quality of life, whereby the children's situation is strongly related to parental coping and capacity. Such parents ask for more help in order to increase their care capacity, while the network is often insecure about how to help and thereby withdraw. They ask for guidance and training to be able to support cancer families. Based on this, the Cancer- Psycho-Educational Program for the SOcial NEtwork (PEPSONE) study was developed.

Objective: To optimize social network support through a psycho-educational program for families living with parental cancer and their network members in order to increase parental capacity and thereby secure the children's safety and quality of life.

Methods: A randomized controlled trial (RCT) in which families (N=60) living with parental cancer will be randomized to either an intervention group or a control group. The intervention will last for 3 hours and includes (1) introduction, (2) psycho-education (living with cancer in the family and the importance of social network support), and (3) discussion (this family's need for social support). Primary outcomes are social support, mental health, and quality of life, and secondary outcomes are resilience and parental capacity. Data will be collected by a set of questionnaires distributed to healthy parents (N=60) living with a partner with cancer, one child in the family between 8-18 years of age (N=60), and network members (N=210) of the intervention families at inclusion, and after 3 and 6 months. Comparing differences between the intervention group (n=30) and the control group (n=30), the power analysis shows that $P<.05$ and a statistical power = .80 would detect effect sizes of clinical interest.

Results: This paper presents the Cancer-PEPSON study's protocol to provide a broader understanding of the background and content of the program. The study is ongoing until August 2016 and the first results are anticipated to be finished by November 2015.

Conclusions: To our knowledge, this will be the first RCT study to optimize social network support through a psycho-educational program for families living with parental cancer and their network members, as well as provide an evidence basis for social network support. The results may provide important knowledge that is useful for clinical practice and further research. The trial is reported according to the CONSORT checklist.

ClinicalTrial: International Standard Randomized Controlled Trial Number (ISRCTN): 15982171; http://www.controlled-trials.com/ISRCTN15982171/15982171 (Archived by WebCite at http://www.webcitation.org/6cg9zunS0)

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KEYWORDS

randomized controlled trial; parental cancer; children; social support; psycho-education; quality of life; mental health

Introduction

Annually, approximately 30,000 Norwegians are diagnosed with cancer. Even if the majority of these are older individuals, more than 3500 children under <18 years of age experience a parent getting cancer. Thus, about 18,000 Norwegian families live with parental cancer [1,2]. Internationally, roughly 14-18% of cancer patients have dependent children, indicating a large population of families for whom cancer poses special challenges [3-5].

Social support is important for human health and quality of life, including emotional, practical and economic help, and information provided to the individual by significant others, such as distant family members, friends, and co-workers, etc [6]. Even if social support represents an essential resource for families living with parental cancer, these parents report a need for more social support and help in order to uphold their parental capacity and to continue a "normal" everyday life [7-9]. Social network members want to support and help, but request support assistance from professionals to provide them with knowledge and various strategies to facilitate better and more prolonged support [7,10]. Based on this, we developed the Cancer-Psycho-Educational Program for the SOcial NEtwork (PEPSONE) study. Cancer-PEPSONE is a randomized controlled trial (RCT) study aimed at optimizing social network support through a psycho-educational program for families and their network members, in order to uphold parental capacity and children's quality of life. This paper presents the study's protocol to provide a broader understanding of the background and content of the study.

Previous Research

In a demanding balance between caring for children, work, and domestic tasks, cancer illness and cancer treatment represent a significant burden and long-lasting strain for the entire family [4,11,12]. These consequences are, however, different for individual family members.

A cancer diagnosis and treatment usually results in multiple consequences for the sick parent, including physical and psychosocial side effects, such as nausea, pain, and fatigue as well as anxiety, depression, and traumatic stress reactions [13,14]. Sick parents are, therefore, frequently challenged in fulfilling their roles in everyday life in relation to work, practical tasks at home, and caring for their children and social life [7,15]. Partners of cancer patients also experience stress reactions, anxiety, depression, and impaired quality of life [16-18]. They frequently report a considerable increase in the strain related to child care, in performing domestic tasks, and in supporting their sick partner as well as being the family's breadwinner. Healthy partners often fulfill double roles and feel distressed, insecure, and lonely [7,11,19]. Together, these challenging strains on both parents may negatively influence their parental capacity and quality of life [12,20,21].

How children are affected by parental cancer depend on the child's age, experiences and maturity, and the cancer severity, but are especially related to parental coping and the family's function in everyday life [4,11,22]. Reduced parental coping negatively affects the children's behavior and their emotional, physical, and school life [23,24]. Young children are mostly affected by concrete changes in daily routines such as frequent hospitalizations and changes in parental behaviors (eg, sadness, fatigue, and impatience) [25]. Older children and adolescents also feel empathy, and they are worried about losing their parent and think about how the cancer will influence their own futures [23,26]. These children are at risk of several physical and psychosocial symptoms such as decreased energy levels, headaches, stomach pain, sleep deprivation, concentration problems, depression, anticipatory grief reactions, and reduced quality of life [19,24]. Additionally, these children report impairments in the family's social life and increased involvement in domestic tasks such as looking after minor siblings [23]. Research indicates that they try to reduce the strain on parents by asking for less help or not bringing friends home, as well as internalizing their own problems and concerns [23,24]. Thus, children living with parental cancer ask for predictability and stability in everyday life, and call for a balance between talking about their current situation and a "space" in which they can talk about things other than cancer, hang out with friends, and participate in leisure activities [7,27].

Several studies have emphasized the importance of social support for physical and mental health, for quality of life, as well as for coping with and recovering from cancer [28-31]. Nonetheless, cancer patients still experience disruptions in their social lives as well as unhelpful help or a lack of social support, especially over time [8,32]. Cancer patients' main concern regards caring for their children, and in particular they want to protect their children and maintain an ordinary everyday life [20,33,34]. A number of these parents express that they are dependent on help from their social network to sustain their regular everyday life, and call for more and prolonged support to uphold their own parental capacity and avoid "hitting the wall" [7,35]. These families often find it difficult to ask for the help and support they actually need and they often experience that help and support drop shortly after the diagnosis and their network withdraws [7-9].

Previous research has found that network members surrounding families in crisis are generally positive in providing support but that they are often insecure about how to help. They report being afraid to say or do the "wrong things", of intruding on the family, or they may assume that their contributions are unimportant or that the family copes adequately on its own [7,8,36]. Network members claim that by acknowledging support efforts and providing various strategies, professionals can facilitate better and more prolonged network support [36].

Most studies of social support related to cancer are descriptive, documenting the importance of social support for physical and psychological health as well as for quality of life [32].

Intervention studies on enhancing social support lag far behind, and are mostly directed to cancer patients, especially breast cancer, focusing on different kinds of support groups [37,38]. Intervention studies related to parental cancer and dependent children are lacking, as are intervention efforts aimed at enhancing social support for children and families members [39]. However, Hogan's review [38] provides support for the overall usefulness of different social support interventions.

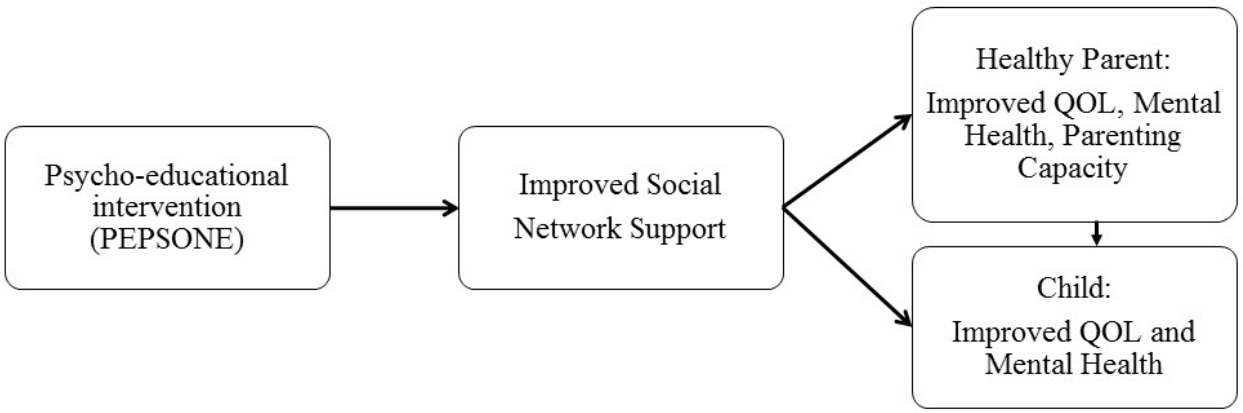
Theoretical Framework

Social support is thought to affect mental and physical health positively through its influence on emotions, cognitions, and behaviors [6,38,40]. Nevertheless, the association between social support, well-being, and health is complex and therefore difficult to conceptualize. House and Kahn's definition of social support makes a conceptual distinction between different types of social support, including emotional, economic and practical help, and the provision of information [41]. Social network support encompasses various kinds of support given to individuals and families by other family members, friends, colleagues, neighbors, and others [6,11]. Over the last decades, much health research has distinguished between "perceived" and "received" social support [6,42]. "Perceived support" refers to the perception by those who are in need of support that such support would be available if needed (ie, qualitative), whereas "received support" refers to the actual support resources received by those in need (ie, quantitative). Two major models have been proposed to explain the link between social support and well-being: the main or direct effect model and the buffering effect model [6,43]. The direct effect model suggests that social support is directly associated with well-being, while the stress buffering model describes how social support can protect individual well-being from the negative impact of stress. Perceived and received support have exhibited different effects on well-being and health, with perceived support demonstrating the most influential effect [6]. Furthermore, social support has been seen as transactional, which means that other factors (eg, personality characteristics, contextual, and interpersonal processes) influence the impact of the support. Nevertheless, exactly what the mechanisms are which provide for the effects of social support remains to be discussed and the causal links are still unclear [6,42].

Cohen [42] argues that strengthening and increasing the availability of support in social networks, and reducing negative interactions within one's network are essential for human health. Psycho-education can be a viable strategy for achieving this. Psycho-education is defined as professionally delivered illness-specific information and tools for managing challenges in everyday life [44]. It builds on a holistic and competence-based approach, focusing on health promotion, collaboration, and empowerment where the development of open communication, competence, knowledge, and skills are crucial elements facilitating behavioral change [44]. Psycho-education looks to support the individual's understanding of a challenging situation and help gain access to resources, develop awareness of issues, foster a sense of control, and educate about coping skills for both families and their networks. These factors focus on improving cognitive awareness and coping skills. However, psycho-education also looks to promote insights that address affective worries and concerns [44]. This study focuses on investigating whether the psycho-education of social networks' members is an appropriate method to achieve beneficial effects with respect to increasing social support and thereby parental quality of life, mental health and parental capacity, and thus the children's well-being. Based on the theoretical framework we have developed a conceptual model of the study (Figure 1).

As shown in Figure 1, it is hypothesized that the Cancer-PEPSONE program will enhance the family's social network support. This enhanced support will have direct effects on healthy parents' quality of life, mental health, and parental capacity, as well as direct and indirect effects on children's quality of life and mental health. The study upholds the child's perspective and complies with the Norwegian Act for Health Personnel [45], putting children's well-being on the agenda when living with parental illness. It also complies with the International Convention on the Rights of the Child (UN), emphasizing the rights of children to care, protection, rehabilitation, and assistance in various situations that can negatively affect their life situation.

Figure 1. Research model.



Objectives and Hypotheses

The purpose of the Cancer-PEPSONE project is to expand the knowledge base and build competence in networks to help children living with parental cancer. The overall aim of the study is to optimize social support from the social network through a psycho-educational intervention. Based on the project's aims and research model, we hypothesize that (1) a psycho-educational program will improve the provisions of social support to the affected family, (2) parental psychosocial health and quality of life will increase through social network support, and (3) the children's psychosocial health and quality of life will improve because of more and better social support and increased parental capacity, mental health, and quality of life.

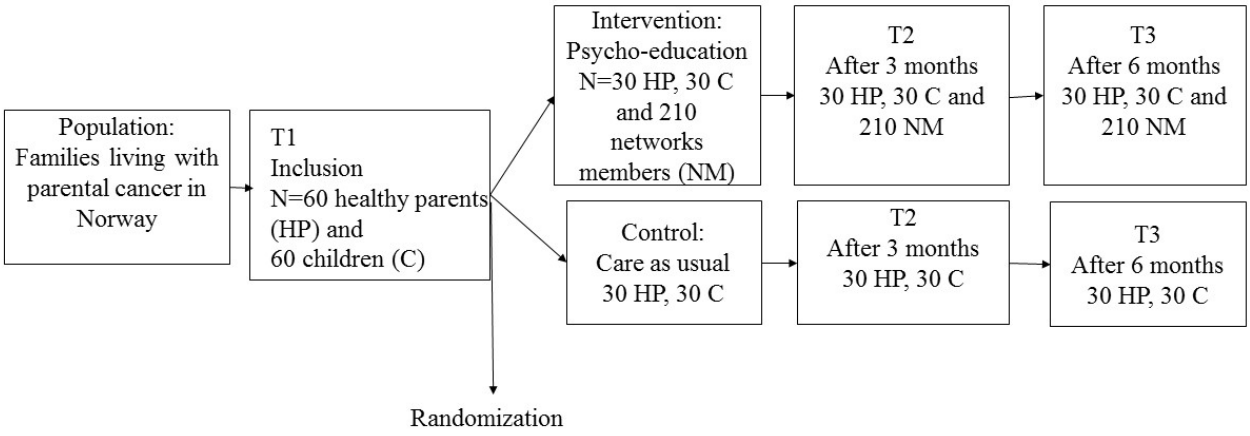
Methods

Trial Design

Based on the study's hypothesis and research questions, the Cancer-PEPSONE study will be conducted as a single center, randomized controlled trial (RCT) [46], including an intervention and a control group. This design is best suited to test the effectiveness of various types of intervention in clinical trials. The Cancer-PEPSONE study design is outlined in Figure 2.

After receiving written and oral information from one of the two first authors, families fulfilling the inclusion criteria are included in the study. The participants receive a form for informed consent and the first set of questionnaires for the healthy parent and one child in the family (T1) by mail. The consent form and the questionnaires are returned to the researchers in pre-stamped envelopes. Then, every other family is randomized to either the treatment group or the control group by one of the researchers based on the order in which the informed consent and questionnaires are returned. The type of intervention makes it impossible to blind the trial participants or the psychologist. The families in the intervention group and their network members receive the intervention, while the control group receives no intervention (care as usual). The control group is offered the intervention after finishing the study participation, after approximately 6 months, and all participating families get a DVD of the program after finishing T3 after 6 months.

Figure 2. Flow diagram of the The Cancer PEPSONE study.



Intervention

The intervention is a psycho-educational program for the family and its network members. The authors developed the program based on clinical experience, previous research, and the theoretical framework previously outlined. It was pilot-tested by two of the authors and thereafter, all the authors discussed and modified the program to its final structure.

The parents in the intervention families decide who in the family and which network members would participate in the program. The program is conducted in the families' homes, or else where they choose, by one of three clinical psychologists, all of whom are experienced in working with families and children in crisis. It lasts for approximately 3 hours. None of the researchers are involved in the intervention that contains the elements described in Textbox 1.

Textbox 1. Elements included in the intervention.

- Welcome and introduction (10-15 minutes): The psychologist leading the meeting introduces herself and presents the content of the program before the participants introduce themselves. The goals of the program are (1) to emphasize the importance of network support; (2) secure social network support for cancer families over time; (3) share knowledge to make each other wiser.
- Psycho-education (approximately 1 hour): In this part, the psychologist has a teaching session focusing on the following areas: (1) the consequences of living with a cancer diagnosis in the family for the sick and the healthy parent, and with a special focus on the children; (2) general reactions and the needs of both the children and the adults; (3) common useful coping strategies for crisis; (4) the importance of social network support, focusing on what is social network support for families in crisis, and what do we know about helpful network support; (5) the importance of "openness" and communication between the family and the network; (6) what can social networks do, and different types of social support (eg, emotional, practical, information, and economical support); (7) how to sustain network support over time: the importance of distribution, and the coordination of support.
- Discussion (approximately 1.5 hours): Based on the teaching session, the goal of the discussion is to enhance the family and its network members' understanding of the value of open communication about the family's need for social support and the network members' ability and willingness to give such support. The psychologist facilitates the discussion based on the experiences from previous support giving/receiving processes between the family and the network, the family's current needs and what is the network able and willing to do as well as coordination of the network support.
- Summing up and closing (10 minutes): The psychologist sums up the main points from the teaching session and the discussion.

A detailed procedure for the intervention is developed and reviewed by the intervention psychologists together with the authors securing that the intervention is performed in the same manner for all families. After the meeting, the psychologists fill out a form with information about how the intervention went according to the protocol, who attended the meeting (roles/relations), the themes discussed, and a short field note to record any observations about the context and impressions arising from the meeting. All participants in the intervention also fill out an evaluation form on how they experienced the psycho-education.

Eligibility Criteria

The study contains 3 samples. Sample 1 and 2 (n=60) consist of 30 families in the intervention group and 30 in the control group. These samples include the healthy parent and one child from each family. The inclusion criteria for these families are (1) a healthy parent having a partner or spouse diagnosed with cancer within the last five years and treated for cancer and (2) one child in every family, aged 8-18 years old, living with a parent who has cancer. With multiple children in the family, the oldest child who is willing to participate will be recruited. The parents in the intervention group ask the number of adult network members (mean 7, limited to 15 network members, N=210) if they want to participate in the intervention. This group of adult network members makes up Sample 3. The inclusion criteria for these network members are (1) extended family members, friends, neighbors, and work colleagues of the parents, (2) ≥18 years, and (3) living nearby the family.

The exclusion criteria for the study are (1) healthy parent not living with the ill parent or the ill parent has died, or having a serious disease, (2) children <8 years old, not living with ill parents, serious disease themselves, and (3) network members living >2 driving hours from the family.

Recruitment

Participants are recruited nationwide using a wide-ranging recruiting strategy including information acquired through hospitals and primary healthcare, brochures, and different websites. Families are also recruited through the Norwegian Cancer Society, the Montebello Cancer Center, child responsible healthcare professionals in hospitals, cancer coordinators in primary healthcare, and resource nurses in cancer care and palliation.

Outcome Measurements and Data Collection

The literature recommends using a range of outcome measures in evaluating complex interventions, as a single outcome may not capture the results or unintended consequences of the study [47]. Therefore, different self-reported questionnaires are included for all participants, where social support, mental health, and quality of life are primary outcomes, while resilience and parent capacity are secondary outcomes.

Outcome Measurements

The questionnaire for the healthy parents includes the information shown in Textbox 2. The questionnaire to the children includes the information shown in Textbox 3. The questionnaire set to the network members includes the information shown in Textbox 4.

Textbox 2. Information included in the questionnaire for the healthy parents.

- Demographic data about the healthy parent: age, gender, education, social status, children, employment status, and income.
- Demographic and medical data about the sick parent: age, gender, type and degree (metastasis) of cancer, months since diagnosis, type of treatment, and months of treatment and current treatment status.
- The Crisis Support Scale (CSS) [48] and the Assistance Questionnaire-Receivers of support (AQR) [49] measure social support. CSS is a short scale for measuring social support after a crisis has occurred, consisting of 7 questions with a rating scale from 1 (never) to 7 (always). All items are summed for a total mean score, where a higher total score indicates more received support. The scale appears to be very robust and to have satisfactory psychometric properties [48]. AQR measures adults' experiences and need for 9 different types of social support related to the situation caused by the cancer. The instrument was developed by Dyregrov et al [49] and is applied on comparable populations both nationally and internationally.
- The Quality of Life Scale (QOLS-N) is used to measure the healthy parent's quality of life [50]: This instrument measures an individual's overall satisfaction with life based on different life domains. It contains 16 items scored on a 7-point Likert scale ranging from "very satisfied" to "very dissatisfied". The items are calculated into the 6 subscales to assess satisfaction with life domains. The dimensions are scored by summing the scores for each item in the subscale. Possible total scores range from 16-112, where a lower score indicates worse quality of life demonstrated satisfactory psychometric properties in several studies [50,51]. The 6 subscales are as follows: (1) physical and material well-being; (2) personal development; (3) relationships with others; (4) participation in social activities; (5) participation in community and civic activities; and (6) recreation.
- The General Health Questionnaire (GHQ-12) is used to assess current mental health and psychological distress, reflecting the inability of normal functioning in regard to distressing experiences [52]. The questionnaire consists of 12 items on a Likert scale ranging from 0 to 3, where a higher sum-score indicates more symptoms of psychological distress and worse mental health. The GHQ-12 is widely used as a reliable screening instrument for psychological distress and minor psychiatric morbidity outside a clinical setting, showing high psychometric properties in various populations [52].
- The Self- Efficacy Parent Task - Short Form (SEPTI-SF) is used to measure 2 dimensions of the parent's self-efficacy, discipline and achievement, showing satisfactory psychometric properties [53]. These dimensions consist of 11 quotes with 6 alternative answers from "highly disagree" (1) to "highly agree" (6). The dimensions are scored by summing the scores for each item in the subscale.
- The Dispositional Resilience Scale-Revised (DRS-15-R/Hardiness) is used to assess the parents' hardiness in meeting challenging life events and situations [54]. The questionnaire consists of 15 quotes with 4 alternative answers scored on a Likert scale ranging from "highly disagree" (0) to "highly agree" (3). The items are summed into the 3 dimensions, commitment, challenge, and control, where a higher score indicate higher hardiness. The instrument is used internationally as well as in Norway, and has demonstrated validity and reliability within a wide range of studies as well as sensitivity to change [54].

Textbox 3. Information included in the questionnaire to the children.

- Demographic data: age, gender, siblings, and grade in school
- The Revised Child Manifest Anxiety Scale (RCMAS) is used to measure anxiety reactions in children [55]. The RCMAS consists of 28 anxiety items and 9 lie (social desirability) yes-or-no items. Sum scores are provided for total anxiety and the 4 sub-scales: worry/oversensitivity, physiological anxiety, social concerns/concentration and a lie scale. A higher score indicates higher levels of anxiety or lie. RCMAS has demonstrated adequate psychometric properties [55].
- Kinder Lebensqualität (KINDL) is used to assess the quality of life of children [56]. KINDL consists of 30 quotes with 4 alternative answers ranging from "never" to "always". The 6 subscales (physical health, emotional well-being, self-esteem, family, friends, and school) are calculated, where higher scores indicate higher QOL. The questionnaire has showed psychometrically acceptable values [56]. KINDL has 3 versions related to age, but since these cover the exact same questions with somewhat different wording, we decided to use Kid-KINDL to cover the entire age-span.

Textbox 4. Information included in the questionnaire to the network.

Questionnaire information

- Demographic data: age, gender, education, social status, work affiliation, and relation to the family/sick parent.
- Assistance Questionnaire: Providers of support (AQP) represents the opposite version of AQR as it measures the social support given.
- GHQ-12, QOLS-N, Hardiness and CSS, as described for the healthy parents.

Data Collection

Healthy parents and the children fill out the entire dataset at inclusion (T1), after 3 months (approximately one months after the intervention for the intervention group), and after 6 months (Figure 2). One of the two first authors provides the families with questionnaires by post and they return them to the researchers in pre-stamped envelopes. The social network members in the intervention group fill out the T1 questionnaires prior to the meeting starting. The network members in the intervention group fill out the same questionnaires after 3 (T2) and 6 months (T3), distributed via the Internet and the website SurveyMonkey.

Power Calculations and Statistical Analyses

When comparing differences between the intervention group (n=30) and the control group (n=30), the power analysis showed that with $P<.05$ and a statistical power = .80, one would be able to detect effect sizes of *t* tests of about 0.65 of one standard deviation or higher [46]. This means an ability of detecting effects sizes of medium size or higher according to Cohen's criteria [46]. This effect level was judged to be of clinical interest, and was within the study's recruitment frame and economy.

All the data will be coded, verified, and entered into IBM SPSS Statistics for Windows Version 22.0. Normality will be assessed through examinations of skewness and kurtosis for all variables. A two-tailed *P* value of <.05 is considered to be statistically significant [46,57]. Descriptive statistics (mean/median or percentages, SD, and ranges) and correlation analyses (Pearson correlation) will be used to describe the data and to explore relationships among them [57]. The outcome variables will provide sum-scores on the interval level. Multiple regression analysis will be used to explore directional relationships among variables and testing the research model. For the estimation of the effect and the relationships of the variables relevant to the intervention, different methods such as *t* tests and structural equation (SEM) analyses will be used [57]. The missing data problem will be analyzed according to the questionnaires' manual. Statistics will be reported in line with the SAMPL guidelines stated by Lang and Altman [58].

Ethical Implications and Risk

The Regional Committee of Research and Ethics in Western (REK West) Norway and the Norwegian Social Science Data Services (NSD) approved the research protocol October 9, 2013 (reference number: 2013/1491/REK vest).

The study will be conducted in compliance with the Declaration of Helsinki [59] and the requirements for data processing outlined in the NSD [60]. Only the two first authors have access to the data files. Procedures for handling data and data security in the study are developed. Study participation is based on written and oral information and written consent [59]. For children <12 years of age, the parents must consent on behalf of the children, whereas for the 12-18 year old children the consent is given from both parts. Study participation is voluntary and participants can withdraw at any time without the provision of reasons or any negative consequences. Serious risks or undesired effects of the intervention or the assessment by questionnaires are not described in the literature and no specific risks related to this study are anticipated. All the professionals participating in the study have extensive experience as researchers or clinicians in the field of working with children, serious illness, crisis, grief, and trauma. This competence will secure ethical and safe conditions for the participants. A referral process for further assistance or treatment for participating families with special needs will be ensured by the psychologist. Any changes in the study protocol will be applied for to REK West and NSD.

Project Organization, Funding, and Timeframe

The Cancer-PEPSONE study is a one-center study conducted in Norway. The study is connected to an interdisciplinary research group and an international advisory board to provide input and secure the quality of the study, independent from the sponsors. The study is fully founded by the Research Council of Norway (4.7 million NOK) and by the Norwegian Directorate of Health (1.3 million NOK). The funders have no role in conducting the study, but reports on progress according to the protocol as well as economy are sent annually.

The study's timeframe is 3 years. It was initiated in August 2013 and will be completed by August 2016. A schematic detailed timeline of the study is outlined in Table 1.

Table 1. Timeline for the Cancer PEPSONE study.

	Study period						
Time point	Autumn 2013	Spring 2014	Autumn 2014	Spring 2015	Autumn 2015	Spring 2016	Autumn 2016
Founding and ethical approval	x						
Advisory board	x	x	x	x	x	x	x
Research group meetings		x	x	x	x	x	x
Recruitment	x	x	x	x			
Enrollment		x	x	x			
Data collection T1		x	x	x			
Data collection T2		x	x	x	x		
Data collection T3		x	x	x	x	x	
Analyzing data					x	x	x
Publishing results						x	x
Study close-out							x

Results

The Cancer PEPSONE study is ongoing, where enrollment of families began in January 2014. As of October 2015, 45 families are enrolled in the study. Of those families, 19 are randomized to the intervention group, with 15 families receiving the intervention together with approximately 120 network members, while 4 families are waiting for the intervention. As well, 20 families are enrolled in the control group and 6 families are not yet randomized. The first results from the study are expected in October 2015.

Discussion

Principal Findings

The objective of this study is to optimize social network support through a psycho-educational program for families living with parental cancer and their network members in order to increase parental capacity and thereby secure the children's safety and quality of life. Until now, the research regarding parental cancer has mainly focused on describing the parents' challenges regarding their risk of impairment of psychosocial health, quality of life, and parental capacity [4,12]. In addition, they have described their positive and negative experiences with social network support [7,8]. Descriptive research documents that children living with parental cancer are especially exposed and vulnerable, and that their physical, psychosocial, and behavioral impairments largely depend upon their parents' coping abilities [23,24]. In previous research, it is documented that it is a match between the bereaved and social networks' accounts of the challenges involved in the support giving and receiving processes [10,36]. However, we lack intervention research to document the effect of systematic programs in order to optimize social support to make families living with parental cancer cope better with their situation. By including a family-, child-, and social network-perspective, the Cancer-PEPSONE study covers an important new area of research both nationally and internationally. Therefore, several important issues underpinning this study are worth highlighting.

The descriptive research has mainly focused on the negative consequences of parental cancer for both the sick and healthy parent as well as for their children. Consequently, these families' experiences are at risk of being pathologized and assessed as being in need of individual professional treatment, instead of assistance in coping and building on their present resources [39]. Acknowledging that the lives for both the parents and the children involve more than just coping with the stress related to cancer, the Cancer-PEPSONE study builds on the inherent resources of family members and their networks, including a requested health promotion perspective into the research [39].

A strength of the Cancer-PEPSONE study is that it originates from the experiences brought forward in clinical practice and through our prior research. It builds upon the cancer families' own outspoken need for more and long-lasting social support, as well as the networks calls for more knowledge and training from professionals as to how to provide good support and help over time [7,10]. The intervention is pilot-tested, securing its usefulness in clinical practice and meeting both the families' and the networks' needs.

Some may question the use of scarce healthcare resources to educate cancer families' social networks. Professionals and researchers have gradually acknowledged that cancer patients live in a social context along with the need to provide psychosocial assistance during cancer more directly and to a greater extent than before [9]. Following legislation, Norwegian healthcare professionals have a statutory responsibility to ensure that the children of seriously ill patients receive information and follow-up during the entire illness trajectory [45]. This implies an obligation to secure psychosocial help for children, either directly or, most preferably, indirectly through parents by increasing their parental care capacity. This perspective is further elaborated in the Norwegian Cancer Strategy 2013-2017 [61], emphasizing a focus on help and support for both children and parents to be able to cope with their situation. These obligations highlight that health service must ensure that families receive assistance with practical, financial, and emotional issues as needed, as well as the municipality's responsibility for coordinating this assistance.

Several families live with cancer over years that often involve irregular changes in the disease trajectory. Adjustment to cancer, therefore, involves a process rather than a singular event. A model that demands extensive professional follow-up over time would be very resource demanding. In contrast, by optimizing the "normal" and available source of social support, the use of formal resources may be limited. As such, limited use of professionals' time spent in educating the families' network members and promoting communication between the family and their social network seems preferable. According to our hypothesis, the psycho-education of the network members will lead to increased confidence in their interactions with friends and families in crisis. Hopefully, this will secure more long-lasting support and prevent "burn-out" in the networks. Most importantly, with more support, the parents may gain more capacity for caring for their children, and thereby prevent the negative consequences of living with parental cancer. Additionally, the improved and increased availability of social support will save society in financial expense.

From a resource perspective, it may also be questioned whether the intervention should be directed towards individual families and their networks, or else to several families and their networks at the same time. We consider this as a crucial element because families living with parental cancer are not a homogeneous group. Each family member faces special challenges and has different needs. Available network resources will also be heterogeneous. In line with this, the Medical Research Council [47] has stated the importance of tailoring complex intervention to local circumstances rather than being completely standardized. By meeting one family at a time, the psychologist can tailor the psycho-education to each family and its network, making sure that their perspective and context is at the basis of the information and conversation. By conducting the intervention in the family's home, the importance of the family's perspective and context is underpinned.

The intervention is four-fold, with an introduction, psycho-education, discussion, and closure. The introduction is important to set the content and the focus of the meeting, and for the participants to briefly get to know the psychologist and

her competence. This allows for building trust and safety within the meeting. One goal of the psycho-education is to provide both the family and its network with knowledge of the specific challenges that adults and children face when living with parental cancer. Even if the family lives in such circumstances, this element can normalize their reactions, being revealing of the other family members' challenges as well drawing focus on what is of most importance for the children. Many families find it difficult to express their own challenges and needs to their networks, and the psycho-education may therefore serve as recognition of their needs in preparing the ground for an open discussion later on. Another main goal of the psycho-education is the focus on the importance of social network support. Again, by providing knowledge to both the family and the network about the complex processes of social support, this may justify the family's needs and give the network some helpful tools to use in the supportive interactions.

Most research has focused on the positive and helpful aspects of social support [6,30]. However, social support can also be experienced as negative or unhelpful, which is an issue that is important to focus on in both the psycho-education and in the discussion part of the intervention. Such negative social support can, for example, be non-helpful advices or advices that the family has not asked for, trivializing of their problems, or that the family feels overrun by help that they do not want or need [37]. In the discussion part, the psychologist builds upon the content of the psycho-education and tailors the content to the actual family and its network, discussing the most relevant aspects based on their situation. A clear goal is to facilitate direct, open and concrete communication between the family and its network members.

A possible limitation of the Cancer-PEPSONE study is that the intervention is short with no follow-up meetings, and therefore that it may be seen as naïve, with little potential to make any changes that last over time. Although the participants in the pilot study would have preferred 1 or 2 repeated meetings, the network members stated that the initial meeting had a good effect in sensitizing them to increased social support. In addition, resource allocation is an important argument. Thus, instead of organizing a follow-up meeting, the psychologists advise and trust the family and its network to organize the network to support themselves. An alternative might be that a local cancer nurse participates in the intervention and then follows up the family thereafter. It may be argued that it is not ethical if no follow-up is done for these vulnerable families. However, they are already enrolled in the public healthcare system, which have legislated obligation regarding follow-up [45]. If the researchers or the psychologists performing the intervention detect special needs of participating families, they will refer them to the public healthcare system. Another limitation of the study might be that we acquire very little knowledge about the qualitative and the procedural aspects of the intervention. However, by including a short evaluation of the program from both the psychologist, the family and the networks' members, including open questions, we acquire more knowledge regarding these aspects. The study is founded as a RTC study. In addition, we will try to fund a qualitative arm of this study, exploring the participants' experiences of both the intervention and its consequences.

The results from this study will be published in at least 6 papers in international peer-reviewed scientific journals, and 3 of these will be included in a PhD thesis. Furthermore, the findings will be presented in chronicles in national newspapers, at conferences, and seminars, both internationally and in Norway. Importantly, if the study results prove successful, we will develop guidelines for the Cancer-PEPSONE to be recommended for the local healthcare system and conducted by public nurses or cancer nurses, for example. The Cancer-PEPSONE program also has the potential to being adapted and studied related to other physical and mental conditions.

Conclusion

There is currently a lack of intervention studies related to parental cancer and children as well as intervention programs enhancing social support between families and social network members. Therefore, the overall aim of the Cancer-PEPSONE study is to optimize social network support through a psycho-educational program for the family and its network members in order to increase the parental capacity and thereby improve the children's quality of life. It is anticipated that the results from this study will support the hypotheses and provide new knowledge about families living with parental cancer. The Cancer-PEPSONE study is innovative given the scope, including a family, a child, and a network perspective, and the intervention and the diversity of measures utilized within this longitudinal RTC design. We hope it will add to the growing body of research on children living with cancer in the family. We anticipate that, based on this study, we will be able to develop a guideline for the Cancer-PEPSONE program. By educating local healthcare professionals or volunteers in using such a guideline, the program is inexpensive and has the potential to be used in other parts of the world as well. By improving network support for these families, less help may also be needed from health professionals.

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Conflicts of Interest

None declared.

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Abbreviations

CSS: Crisis Support Scale
GHQ-12: General Health Questionnaire
NSD: Norwegian Social Science Data Services
PEPSONE: Psycho-Educational Program for the SOcial NEtwork
QOLS-N: Quality of Life Scale
RCT: randomized controlled trial
REK West: Regional Committee of Research and Ethics in Western Norway

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Original Paper

Guided Internet-Based Parent Training for Challenging Behavior in Children With Fetal Alcohol Spectrum Disorder (Strongest Families FASD): Study Protocol for a Randomized Controlled Trial

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Abstract

Background: Fetal alcohol spectrum disorder (FASD) is a term used to encompass the full range of neurobehavioral and cognitive dysfunction that may occur as a consequence of prenatal alcohol exposure. There is relatively little research on intervention strategies that specifically target the behavioral problems of children with FASD. Availability and access to services are barriers to timely and effective care for families. The Strongest Families FASD intervention was recently adapted from the Strongest Families "Parenting the Active Child" program to include FASD-specific content delivered via an Internet-based application in conjunction with 11 telephone coaching sessions.

Objective: Our objectives are to (1) evaluate the effectiveness of Strongest Families FASD in reducing externalizing problems (primary outcome), internalizing problems, and parent distress (secondary outcomes) in children aged between 4 and 12 years diagnosed with FASD when compared to a control group with access to a static resource Web page; (2) evaluate the effectiveness of Strongest Families FASD in improving social competence (secondary outcome) in school-aged children aged between 6 and 12 diagnosed with FASD when compared with an online psychoeducation control; and (3) explore parental satisfaction with the Strongest Families FASD online parenting program.

Methods: Parents and caregivers (N=200) of children diagnosed with FASD who have significant behavioral challenges, ages 4-12, are being recruited into a 2-arm randomized trial. The trial is designed to evaluate the effectiveness of the Web-based Strongest Families FASD parenting intervention on child behavior and caregiver distress, compared to a control group receiving access to a static resource Web page (ie, a list of FASD-specific websites, readings, videos, and organizations).

Results: The primary outcome will be externalizing problems measured by the Child Behavior Checklist (CBCL). Secondary outcomes include (1) internalizing problems and (2) social competence, both measured by the CBCL; and (3) parental distress measured by the Depression Anxiety Stress Scale-21. The Client Satisfaction Questionnaire-8 (CSQ-8) and the Satisfaction

Survey are completed by the intervention group at the end of session 11. Results will be reported using the standards set out in the Consolidated Standards of Reporting Trials (CONSORT) Statement.

Conclusions: It is hypothesized that the Strongest Families FASD intervention group will improve child behavior and parental distress. Caregiver satisfaction is anticipated to be positive. Advancing evidence on the effectiveness and acceptance of distance services can inform policy and adoption of eHealth programs.

ClinicalTrial: ClinicalTrials.gov NCT02210455; https://clinicaltrials.gov/ct2/show/NCT02210455 (Archived by WebCite at http://www.webcitation.org/6bbW5BSsT)

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KEYWORDS

fetal alcohol spectrum disorder; neurobehavioral disorder; prenatal alcohol exposure; disruptive behavior; children; Strongest Families; parenting; randomized controlled trial; eHealth; Web-based intervention

Introduction

Prenatal alcohol (ethanol) exposure is the leading known cause of developmental disability in Canada and is the most prevalent preventable cause of congenital neurobehavioral dysfunction in the Western world [1]. Despite attempts to increase public awareness of the risks associated with drinking during pregnancy, a report from the Public Health Agency of Canada indicates that a significant proportion (10.5%) of pregnancies in Canada are alcohol exposed [2]. The term fetal alcohol syndrome (FAS) was introduced over 40 years ago [3-5] as a diagnosis for children who exhibit the triad of central nervous system (CNS) dysfunction, growth deficiency, and characteristic craniofacial dysmorphology resulting from maternal consumption of alcohol during pregnancy. Of these features, it is the CNS injury that is most debilitating and can manifest as intellectual, neurological, and behavioral abnormalities. More recently, the term fetal alcohol spectrum disorder (FASD) was established to encompass the full spectrum of teratogenic effects induced by ethanol [6]. FAS is believed to occur in approximately 1 to 3 per 1000 live births in North America, and it is estimated that FASD may occur as frequently as 1 in 100 live births [7]. Moreover, recent epidemiological studies suggest prevalence rates as high as 2%-5% [8]. In addition, the total adjusted annual cost associated with FASD in Canada was estimated at US $5.3 billion [9]. The substantial personal impacts of FASD to Canadian families coupled with the economic costs illustrate the need for evidence-based prevention and early intervention.

A key goal of the Public Health Agency of Canada's FASD initiative is to improve the outcomes for people living with FASD. The agency does this by developing practical resources and tools for professionals that build understanding and health professional capacity. These resources complement the substantial work undertaken by provincial and territorial governments to increase diagnostic capacity for FASD [7]. Despite these improvements, there remain challenges with the initial identification of individuals who may be potentially affected by FASD, as well as significant logistical issues, including access to diagnosis in rural and isolated communities.

Once diagnosed, the need for services and supports for children and their families remains largely unmet [10]. Parents and caregivers (henceforth referred to as "parents") of children with FASD are often confronted with significant behavioral challenges without adequate resources and information to help them manage these symptoms [11]. Despite a variety of psychosocial interventions aimed at supporting individuals with neurobehavioral disorders, relatively little research exists that is specifically aimed at improving the behavioral challenges associated with FASD [12]. Problems with service delivery are further compounded in smaller centers and in rural and isolated communities where access to specialized support programs is extremely limited. Even when services are available, significant barriers—such as the costs associated with traveling to clinics for repeated consultations and time spent away from work—make it difficult for families to benefit from them. Additionally, frequent turnover of program personnel often leads to uneven quality and consistency of program delivery. Carefully designed and implemented alternative service delivery models have the potential to meet the needs of children and families with FASD where traditional models have shown difficulty.

The Strongest Families FASD Parenting Program is adapted from the evidence-based Strongest Families "Parenting the Active Child Program" [13-15], which was designed to address many of these issues by providing distance services to families with children exhibiting behavior problems. Program content from the Strongest Families Institute (SFI), a not-for-profit that provides a number of distance-based psychosocial interventions to children and families, was revised to include information specific to FASD, relevant examples, and modification of some curriculum skills. The content is housed within Intelligent Research and Intervention Software (IRIS) [16], a secure Web-based platform that supports interactive and persuasive system design features (eg, personalization, reminders). Via IRIS, parents work online through a progressive skill-based curriculum that includes interactive exercises, instructional videos, and audio clips. Coaches are able to log in to IRIS to see the activities the parent has completed, facilitating their role of supporting parents throughout the program. Telephone coaching sessions are scheduled at convenient times for the parent, thereby removing barriers to care and providing services in the comfort and privacy of their own homes. Participants receive study emails via a secure inbox within IRIS.

Objectives

Our first objective is to evaluate the effectiveness of the Strongest Families FASD program in reducing externalizing problems (primary outcome), internalizing problems, and parent distress (secondary outcomes) in children aged between 4 and 12 years old diagnosed with FASD as compared to a control group with access to a static resource Web page.

We hypothesize that the effects of the Strongest Families FASD intervention will be to reduce the child's externalizing and internalizing problems (as measured by the Child Behavior Checklist (CBCL) externalizing and internalizing scales) and reduce parental distress (as measured by the Depression Anxiety and Stress Scale-Short Form, DASS-21), compared to the control group.

Our second objective is to evaluate the effectiveness of the Strongest Families FASD program in improving social competence (secondary outcome) in school-aged children between 6 and 12 years of age diagnosed with FASD as compared to an online psychoeducation control.

We hypothesize that the effects of the Strongest Families FASD intervention will be to improve the child's social competence as measured by CBCL Social Competence Scale compared to that for the control group.

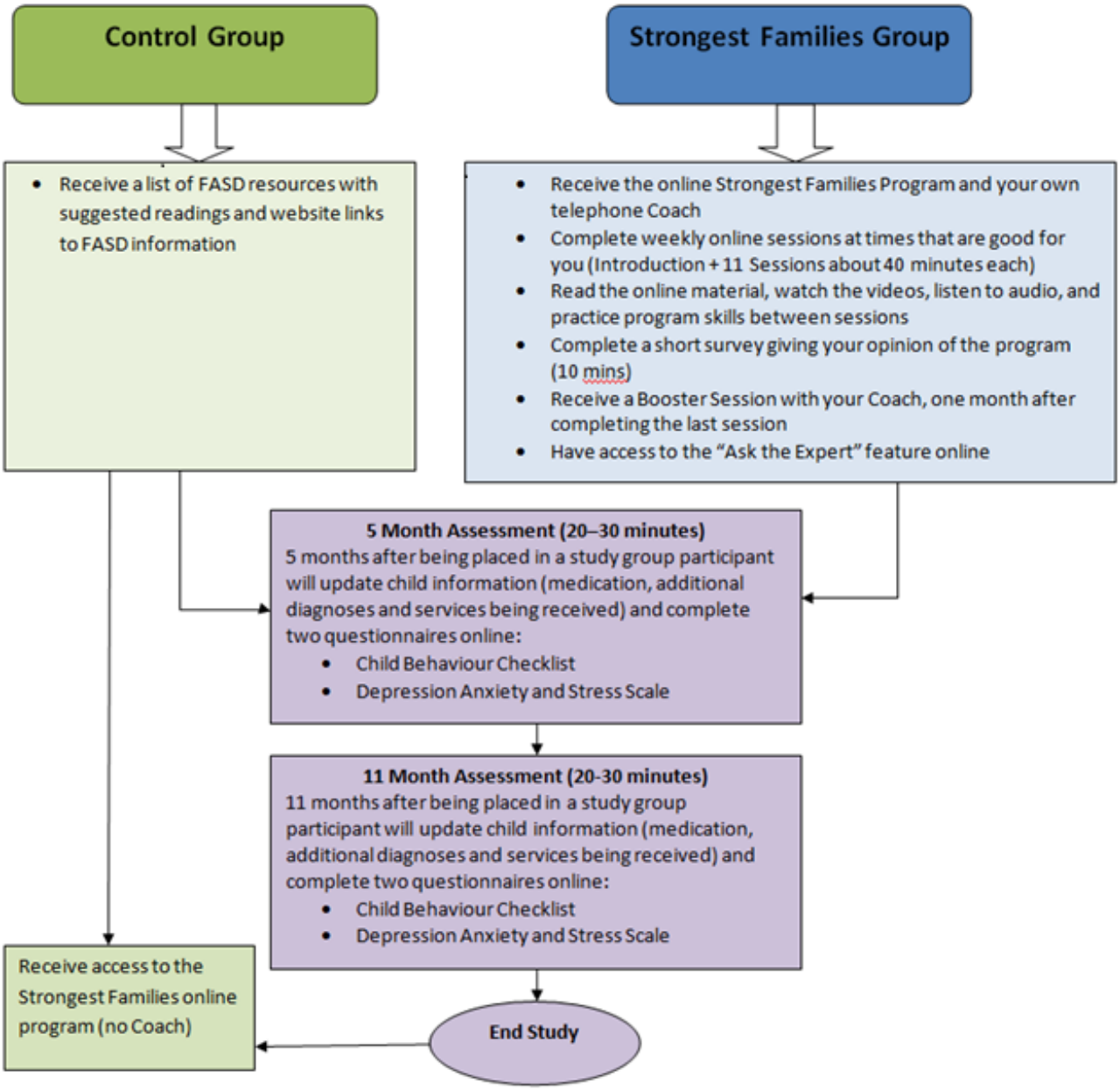
Our final objective is to explore parental satisfaction with the Strongest Families FASD online parenting program.

Methods

Study Design

This study is a 2-arm randomized controlled trial comparing participants assigned to receive either the Strongest Families FASD Web-based parent-training program (SF Intervention Group) or a static resource Web page (Control Group). Neither group will be restricted from accessing additional programs or services during study participation. The Consolidated Standards of Reporting Trials (CONSORT) recommendations [17] are used to guide the methodology. Details of the study design are illustrated in Figure 1. The study has been approved by the Queen's University Health Sciences & Affiliated Teaching Hospitals and IWK Health Centre Research Ethics Boards. The primary and secondary outcome measures will be administered to all participants at baseline (pre-randomization), 5 months, and 11 months post-randomization. The Client Satisfaction Questionnaire (CSQ-8) and Strongest Families Program Satisfaction Questionnaire: FASD Version will be administered to the SF Intervention Group upon completion of the program. Study participation ends for participants in both study groups after completing the 11-Month Follow-up Assessment.

Figure 1. Study flow.



Measures

Child Behavior Checklist

The CBCL is a standardized questionnaire that assesses a child's adaptive functioning and behavior problems by asking parents to rate the frequency with which their child displays each of a series of behaviors. Most items are measured on a 3-point Likert-scale, where 0 means "Not True (as far as you know)" and 2 means "Very True or Often True." Scores are then summed across a number of different subscales and compared to a normative sample to provide a *t*-score for each subscale indicating if the child's behavior is within "normal" limits, "borderline clinical," or "clinical." It is demonstrably sensitive to the effects of parent training programs [18,19]. We will use 2 parent-report versions of the CBCL—the preschool version (CBCL/1½-5) [20] and the school-age version (CBCL/6-18) [21]—to accommodate the age range of the sample. The CBCL requires 15-20 minutes to complete.

CBCL/1½-5

The CBCL/1½-5 consists of 7 syndrome scales (eg, Emotionally Reactive, Anxious/Depressed, Somatic Complaints, Withdrawn, Sleep Problems, Attention Problems, and Aggressive Behavior). Attention Problems and Aggressive Behavior items are grouped into an Externalizing Factor. The remaining scales, with the exception of Sleep Problems, are grouped into an Internalizing Factor. Overall test-retest reliability over 2 test periods averaging 8 days is reported as r= 0.85. Reliability for the 24-item Externalizing and 36-item Internalizing Scales is reported at alpha=.87 and alpha=.90, respectively. Internal consistency is reported for the Externalizing Scale at alpha=.92 and for the Internalizing scale at alpha=.89 [20].

CBCL/6-18

In addition to asking parents to rate frequency of their child's behaviors, the CBCL/6-18 asks parents to provide information about their child's participation in academic, social, and extracurricular activities. The CBCL/6-18 consists of 8 syndrome scales (eg, Anxious/Depressed, Withdrawn/Depressed, Somatic Complaints, Social Problems, Thought Problems, Attention Problems, Rule-Breaking Behavior, and Aggressive Behavior), which group into 2 higher order factors: Internalizing and Externalizing behaviors. This version of the CBCL also provides competence scales for activities, social relationships, and school performance and a total competence score. Overall 8-day test-retest reliability was r=0.92. Reliability for the 35-item Externalizing and 32-item Internalizing scales are alpha=.92 and alpha=.91, respectively. Internal consistency for the Externalizing and Internalizing scales are alpha=.94 and alpha=.90, respectively [21].

Depression Anxiety and Stress Scale-Short Form

The DASS-21 [22] will be used to evaluate parent distress. The DASS-21 is a 21-item self-report measure that asks participants to indicate the extent to which they have experienced a series of mental health symptoms in the past week, ranging from 0 (did not apply to me at all) to 3 (applied to me very much or most of the time). Items belong to 1 of 3 subscales (eg, depression, anxiety, and stress) that can be combined into a composite measure of general distress [23]. The DASS-21 demonstrates strong internal consistency with alpha values at .84 for anxiety, .90 for stress and .91 for depression [22]. The DASS-21 has proven sensitive to the effects of parent-focused interventions [24] and requires 5-10 minutes to complete.

Client Satisfaction Questionnaire

The CSQ-8 has been widely used in primary care and mental health treatment to measure patient/client satisfaction with services received [25]. The CSQ-8 consists of 8 items asking patients/clients to rate the services on a 4-point Likert scale. An overall score for the CQS-8 is produced by summing all item responses. Scores range from 8 to 32, with higher values indicating higher satisfaction. Internal consistency for the CSQ-8 is reported with alphas ranging from .83 to .93 [26]. Participants (SF Intervention Group only) will be asked to rate the quality of service they received as part of the Strongest Families FASD Program.

Strongest Families Program Satisfaction Questionnaire: FASD Version

The in-house developed Strongest Families Program Satisfaction Questionnaire: FASD Version asks participants to rate their agreement with statements about the Strongest Families Program specific to coaching, program components (eg, written materials, videos, etc), and the website on a 5-point scale (strongly agree to strongly disagree). A lower score indicates higher satisfaction. The psychometric properties of this tool have not been tested.

Sample

A sample of 200 parents of children with FASD, aged 4-12 years, will be recruited into the study. Our sample size estimate was based on the minimal clinically important difference in change in outcomes from 0 to 5 months. We have expressed this effect size as a moderate (d=0.50; ie, one half a standard deviation) difference in reduction on CBCL externalizing score for intervention group compared to control. Setting our Type I error rate (alpha) at 0.05. Thus, we require 85 participants in each group for a power of 0.90 and a total sample size of 170 (we will recruit 200 to account for losses). It is reasonable to expect this effect given the larger effects seen in children with oppositional defiant disorder and attention-deficit hyperactivity disorder [12]. The age range for the sample was chosen because interventions provided at this stage can help to prevent the development of secondary problems and because parent-training methods have been most highly developed for this age group [27]. The target sample size is attainable based on the strong relationships we have with FASD diagnostic clinics and FASD support groups across Canada.

Recruitment

The recruitment strategy for this study will be broadly based across Canada. Advertisement materials will include posters, brochures, Web ads, promotional videos, and social media postings. Social media posts (eg, Facebook, Twitter, LinkedIn) will be used to increase study visibility and reach our target sample. Text for social media posts will change frequently to maintain interest in the study, encourage "sharing," and extend recruitment reach. Also, our collaborators at Queen's University have informed former study participants, who agreed to be contacted for future research opportunities, about this study. Partnerships with FASD clinics and support programs across Canada have been established and maintained with study progress updates. The study team promotes the study through conference presentations and exhibits. All interested individuals are directed to a study recruitment website (myStudies) to receive study information, screen for eligibility, and complete online consent (if eligible). The use of myStudies as a medium for screening and consent has been approved by the IWK Research Ethics Board.

Eligibility Screening (Phase 1)

Eligibility screening is a 2-step process that involves a preliminary online screening and then a more comprehensive screening process.

Phase 1 preliminary online screening is conducted on the myStudies website, where potential participants will be invited to complete screening questions for eligibility. Individuals may request (via the website) contact with study staff if they would like to speak to someone about the study. Primary caregivers who are not legal guardians (eg, foster parents, relatives) of the child to whom the study pertains will be required to obtain authorization from the child's legal guardian before taking part in the study. Answers to the screening questions will be automatically assessed in myStudies using predefined acceptance criteria and individuals will immediately receive an on-screen message stating whether they are eligible or ineligible to continue to the next step. Individuals who are eligible at screening will be invited to proceed to an online consent process, also hosted on the myStudies site. Individuals who are ineligible will be informed that they do not meet the study requirements,

thanked for their interest in the study, and encouraged to take part in future studies.

Parents must meet all of the following criteria to be eligible to participate in the study:

1. Have a child between 4-12 years of age with a diagnosis under the umbrella term Fetal Alcohol Spectrum Disorder (as reported by parents) who has been experiencing behavioral problems (as defined by the parent) for at least 6 months prior to study screening;
2. Have been the primary caregiver for a minimum of 6 months prior to entry into the study;
3. Have a reasonable expectation of being be the primary caregiver for at least 6 months after study enrolment;
4. Have the ability to read, write, and understand English;
5. Have access to a telephone;
6. Have access to a computer connected to the Internet; and
7. Live in Canada.

Endorsement of any of the following criteria will exclude parents from taking part in the study:

1. Child is not able to speak in full sentences or understand everyday language and instructions from parent;
2. Parent or child has been diagnosed with psychosis;
3. Child has been diagnosed with schizophrenia, bipolar disorder, or major depression;
4. Child has been identified as putting others at risk of serious harm (ie, requiring medical attention);
5. Parent has taken part in Triple P, COPE, or Incredible Years parenting program within 6 months of completing the study screening; or
6. Parent has previously taken part in a Strongest Families Parenting Program.

The nature of the intervention program necessitates exclusion criterion 1 to ensure that any issues with developmental/cognitive functioning will not impact the child's ability to respond to basic parental instructions related to program skills. Exclusion criteria 2, 3, and 4 are designed to safeguard against the risk of enrolling individuals with potentially complex needs for which the program or study is not appropriate. Criteria 5 and 6 pertain to the parents' experience with programs that teach similar skills.

Eligibility Screening (Phase 2)

Phase 2 screening involves completion of the CBCL baseline measure to determine eligibility for randomization. DASS-21 scores will not impact eligibility. All participants who complete the baseline assessment receive a US $25 gift card by mail or email (their choice).

The following criteria must be met in order for participants to be randomized into a study group:

1. CBCL Externalizing t-Score ≥64 (clinical range);
2. Parental report of a formal diagnosis of one of the following disorders captured under the umbrella term FASD: Fetal Alcohol Syndrome (FAS); partial fetal alcohol syndrome; alcohol related neurodevelopmental disorder; static encephalopathy (alcohol exposed); neurobehavioral disorder (alcohol exposed); and other (To determine possible valid diagnoses not included on our predefined list, parents may report an "other" diagnosis that they believe falls under the umbrella term FASD. These diagnoses will be confirmed by the co-principal investigator on a case-by-case basis and will be accepted if the diagnosis identified (a) has been provided by a recognized FASD diagnostic clinic and (b) conforms to one of the recognized diagnostic schemes used in the assessment of children suspected of having an FASD.);
3. Meets the following criteria for behavior suggestive of FASD adapted from the Neurobehavioral Screening Tool (NST) [28,29]: (a) For children age 6-12 (CBCL/6-18), participants must endorse 7 out of 8 of the items listed in Table 1; or (b) For children age 4-5 (CBCL/1½-5), participants must endorse 3 out of 5 of the items listed in Table 1 with at least 2 endorsements from the items marked with an asterisk.
4. No suicide attempts within the previous 6 months for children 6-12 years of age (screened for using the CBCL/6-18 only); and
5. No current risk of suicide attempts for children 6-12 years of age (screened for using the CBCL/6-18 only).

Table 1. Parent endorsements required to meet clinical eligibility criteria.

CBCL Item	CBCL/1½-5	CBCL/6-18
Acts young for age	✓	✓
Can't concentrate or poor attention*	✓	✓
Can't sit still, restless, hyperactive*	✓	✓
Disobedient at home*	✓	✓
Doesn't show guilt after misbehaving	✓	✓
Argues a lot		✓
Impulsive or acts without thinking		✓
Lying or cheating		✓

Endorsement of CBCL/6-18 item 18 (deliberately harms self or attempts suicide) will generate automatic probes for details within IRIS. Endorsement of current suicide risk for a child who is not being monitored by a health care professional will prompt an automatic onscreen recommendation to seek professional help. This recommendation will be re-iterated to

the participant (parent) in the Assessments Results Letter and followed up with a phone call from study staff.

Randomization and Allocation

Random allocation to the intervention or control group in 1:1 ratio was established independently by an external researcher using a permuted blocked randomization procedure. The external researcher concealed the group placements in sequentially numbered, sealed double envelopes to maintain blinding of study staff. Envelopes are opened sequentially by the coordinator (or delegate) and only after the enrolled participants complete the baseline assessments and are deemed eligible.

Intervention Group

The online Strongest Families FASD program is provided to the parent with weekly telephone coaching sessions; there is no contact with the child. The program consists of evidence-based parenting strategies provided in 11 online sessions using easy-to-read text, demonstrative video and audio clips, interactive questionnaires, and practice exercises. Participants enter information (eg, child's name and specific behavior problems) into IRIS to customize the program content. The weekly coaching sessions facilitate skill acquisition and successful implementation using problem-solving and role-playing techniques. Supplementary program materials (eg, Reward Chart, a Daily Strengths Chart, a Visual Schedule Template, Tryout Pages, and Tips for Teachers Information Sheet) are sent to participants via mail. These materials are provided in physical format to enable parents to post the materials in a convenient place in the home and to facilitate communication between home and school or day care. A booster telephone coaching session is offered 1 month after completion of the program to check in with the parent to review consistent use of the skills, prevent or deal with setbacks, and provide support and encouragement, if needed. An experienced coach supervisor will conduct weekly case review with coaches to problem-solve and to review quality assurance and adherence to protocols.

The effectiveness of the principal components of the intervention has been supported in previous studies [13-15] and are currently being used by the SFI Service Program. The content was revised to tailor the intervention to the needs and challenges specific to families affected by FASD. Changes were informed by data collected in a series of telephone interviews with families and clinicians who have personal and professional interest and expertise in the field [30].

An "Ask the Experts" message board feature within IRIS allows parents to receive answers to individual questions from 1 of 2 designated experts with extensive clinical and research experience within the FASD population. A question deemed by the Principal Investigator (or delegate) to be of potential interest to the larger group will be de-identified and posted to the message board with a response. The author of that question will be notified by email and directed to the message board to view the response. Questions that are not appropriate or deemed not of potential interest for the larger group will be answered individually via email.

Participants receive progress reports midway through and at the end of the program, summarizing parent ratings of their child's improvement across sessions since the beginning of the program. The parent submits ratings at the beginning of the next online session (eg, parent rates Session 1 at the beginning of Session 2) so that they have time to practice the previous skill and evaluate their child's improvement.

Participants who do not maintain regular contact with their coaches will be sent a Re-Contact Letter via email after three consecutive missed phone calls followed by 5 days without response to an email from the coach. The file will be placed on temporary hold until the participant re-establishes contact or closed at the end of the projected study period. Automatic email reminders will be sent to the participant approximately every 6 weeks, inviting them to resume the program or to complete assessments.

Control Group

The Control Group will receive access to a static (ie, noninteractive) FASD resource Web page within IRIS that provides a list of recommended book titles, videos, websites, and organizations.

Participants in the Control Group receive a Mid-Study Progress Message approximately 10 weeks after randomization (coinciding with the SF Intervention Group's mid-program progress report). The message thanks participants for taking part in the study, encourages them to visit the static resource page, and reminds them of the 5-Month Follow-up Assessment.

Participants in the Control Group will gain access to the Strongest Families FASD program (without coaching) after their study participation is complete. The program is designed to be self-directed and contains built-in tutorials to help users navigate the website, though technical support is available. The Strongest Families program will be accessible until 6 months after the last Control Group participant receives access to the website.

Follow Up Assessments

Participants will complete follow-up assessments at 5 and 11 months post-randomization. Participants in both study groups will receive an email message at 145 days (for the 5-month assessment) and 325 days (for the 11-month assessment) prompting them to complete the appropriate follow-up assessments online within IRIS. The assessments will take approximately 20-30 minutes in total to complete. Participants will be asked to complete the DASS-21 and the CBCL and to update some demographic information (eg, child's medications, any new diagnoses). Up to 3 email reminders will be sent and a courtesy phone call reminder will be made for incomplete assessments approaching their expiry date. For each completed follow-up assessment, participants will receive a US $25 gift card by mail or online and a summary of their assessment results via email (within IRIS).

Assessment Results Letters (baseline and follow-ups) will report if the child's CBCL scores fall within the clinical range for the Anxious/Depressed (CBCL 1½-5 & 6-18), Withdrawn/Depressed (CBCL/6-18), or Withdrawn

(CBCL/1½-5) scales and will also include the parent's depression, anxiety, and stress scores (DASS-21) as compared to the general population (ie, about the same, higher, or much higher than most people). Additionally, the child's Externalizing CBCL scores will be reported as being the same, improved, worse, or resolved (within the normal range) as compared to baseline. Letters will emphasize to the participant that assessment results are not a diagnosis and do not replace medical care. Participants will be able to request a phone call from study staff to discuss assessment results.

Statistical Analyses

Data will be analyzed by a statistician blinded to group assignment. The study design is a 2 (group: intervention vs control) × 3 (time: baseline, 5 months, 11 months) mixed factorial with repeated measures.

An intention-to-treat analysis will be performed using a full information maximum likelihood [31] mixed-effects regression framework. Specifically, for the primary outcome of the study we will create a hierarchical ("stacked") data set and regress the CBCL externalizing scores on group (coded as Control=0 and Intervention=1), time (coded naturally as baseline=0, 5 months=5, etc) and the group × time interaction. Child age, sex, comorbidities, and medications have been identified a priori as variables to be added to the model. The critical test will be the group × time interaction. Based on the described coding, the parameter for this effect will be the estimated differential change on the CBCL externalizing score between the control and intervention groups per month. The overall effect will be this parameter estimate × 5. We anticipate using an unstructured covariance matrix for deriving the error term.

Discussion

Research has identified significant gaps in the capacity to treat FASD [10,11]. Continued evaluation of services established to provide support for families affected by FASD will highlight where gaps in care persist and provide evidence to modify services to obtain best practices. Intervention programs that support children and their families by strengthening their home environment and support systems are critical to maximizing the children's potential and modifying secondary effects [12]. Reviews in this area have recognized that the specific challenges experienced by families of children with FASD can vary significantly and, consequently, each individual requires a personalized management program [32]. The Strongest Families FASD Parenting Program, adapted from the evidence-based Strongest Families "Parenting the Active Child Program" [13-15], is designed to address many of these issues by providing personalized distance services to families with children exhibiting behavior problems.

Conclusions

Recruitment began July 2014. This study will (1) evaluate the effectiveness of the Strongest Families FASD program in reducing behavior problems and improving caregiver stress; (2) determine the feasibility of the Strongest Families online FASD Parenting Program for caregivers; and (3) inform eHealth service delivery policy and potentially influence uptake of the Strongest Families-FASD Program. The ultimate goal is to improve timely access to evidence-based eHealth services for families living with FASD.

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Conflicts of Interest

Strongest Families Institute is a not-for-profit organization delivering services to Canadian families. Dr. McGrath is co-founder and chair of the board of SFI; he derives no financial benefit from SFI. Dr. Lingley-Pottie is co-founder, president, and chief executive officer of SFI; she provides her academic and clinical consultation as an in-kind contribution to this study. Dr. Lingley-Pottie will be a co-founder of the IRIS company that is pending incorporation and may benefit financially in the future from IRIS and services sales.

Multimedia Appendix 1

Grant Funding Agency (Canadian Institutes of Health Research) Peer Review.

[PDF File (Adobe PDF File), 215KB - resprot_v4i4e112_app1.pdf]

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Abbreviations

CBCL: Child Behavior Checklist
CSQ-8: Client Satisfaction Questionnaire
DASS: Depression Anxiety and Stress Scale
FASD: fetal alcohol spectrum disorder
IRIS: Intelligent Research and Intervention Software
SFI: Strongest Families Institute

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Original Paper

Games and Telerehabilitation for Balance Impairments and Gaze Dysfunction: Protocol of a Randomized Controlled Trial

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Abstract

Background: Digital media and gaming have received considerable interest from researchers and clinicians as a model for learning a broad range of complex tasks and facilitating the transfer of skills to daily life. These emerging rehabilitation technologies have the potential to improve clinical outcomes and patient participation because they are engaging, motivating, and accessible. Our research goal is to develop preventative and therapeutic point-of-care eHealth applications that will lead to equivalent or better long-term health outcomes and health care costs than existing programs. We have produced a novel computer-aided tele-rehabilitation platform that combines computer game-based exercises with tele-monitoring.

Objective: Compare the therapeutic effectiveness of an in-home, game-based rehabilitation program (GRP) to standard care delivered in an outpatient physical therapy clinic on measures of balance, gaze control, dizziness, and health-related quality of life.

Methods: A randomized, controlled, single-blind pilot trial will be conducted. Fifty-six participants with a diagnosis of peripheral vestibular disorder will be randomly assigned to either usual physical therapy (comparator group) or to a game-based intervention (experimental group). Measures to be assessed will include gaze control, dynamic balance, and self-reported measures of dizziness.

Results: The project was funded and enrollment was started in August 2014. To date, 36 participants have been enrolled. There have been 6 drop-outs. It is expected that the study will be completed January 2016 and the first results are expected to be submitted for publication in Spring of 2016.

Conclusions: A successful application of this rehabilitation program would help streamline rehabilitation services, leverage therapist time spent with clients, and permit regular practice times at the client's convenience.

Trial Registration: Clinicaltrials.gov: NCT02134444; https://clinicaltrials.gov/ct2/show/NCT02134444 (Archived by WebCite at http://www.webcitation.org/6cE18bqqY)

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KEYWORDS

balance-exercises; gaze-exercises; home therapy; telerehabilitation; therapeutic-gaming; vestibular rehabilitation

Introduction

The vestibular sense organs of the inner ear are required to stabilize gaze during head motion and provide an absolute frame of reference with respect to gravity for body orientation and balance. Damage to the vestibular sense organs can lead to a number of symptoms and functional difficulties, including blurred vision, dizziness, disorientation, and falls [1]. The economic impact of vertigo and chronic imbalance is immense. Statistics from 2001 to 2004 estimated that 69 million people in the Unites States aged 40 and older report vertigo and imbalance [2-4].

The recovery of function that patients show following the damage due to peripheral vestibular lesion has long been recognized. Following peripheral vestibular lesions, sensory deprivation studies show minimal recovery [5], whereas targeted activities in enriched environments demonstrate considerable recovery of function [6-9]. A recent meta-analysis [10] demonstrated a moderate to strong evidence that a task-specific approach to vestibular rehabilitation is an effective intervention for unilateral peripheral vestibular dysfunction (PVD).

Improved patient compliance with rehabilitative care plans and increased access to these health services can improve health outcomes for individuals with chronic disabilities. In this regard, recent studies provide descriptions of the benefits of activities facilitated through video gaming. For example, studies have used the Nintendo WiiFit balance board to train and assess balance in elderly patients [11-13]. A similar approach has been developed by Szturm and colleagues [14-16] and has extended balance gaming exercises to more demanding surfaces. A recent case study of 3 individuals with PVD by Po-Yin Chen et al [17] involved adapting the Wii mote game controller to track head motion, and to use quick head movements to trigger events in a custom computer game (ie, to initiate a baseball bat to swing and hit a baseball). A preintervention and postintervention cases series was recently conducted by Szturm and colleagues [18] in 9 adults diagnosed with PVD who received a home game-based vestibular exercise program. They showed the feasibility and usability of the program. Their findings also demonstrated that using head rotation to interact with computer games, when coupled with demanding balance conditions, resulted in substantial improvements in gaze control, standing balance, and stability of the upper trunk during walking. According to the Dizziness Handicap Inventory (DHI), a self-rating, 25-item questionnaire used to quantify a participant's perception of dizziness and its impact on quality of life, perception of dizziness decreased significantly. These findings provide preliminary support that a low-cost home game-based exercise program is well-suited to train gaze-control and balance.

Methods

New telerehabilitation technologies (ie, digital media and innovative computer input devices) may improve clinical outcomes by making rehabilitation more motivating, accessible, and ecological. The research objectives of this study are

1. Compare the therapeutic effectiveness of a home game-based rehabilitation program to standard care delivered in an outpatient physical therapy clinic on measures of balance, gaze control, dizziness, and health-related quality of life; and

2. Examine the time course of change electronic gaze performance measures of participants in the home program.

It is postulated that the home game-based rehabilitation program will result in greater improvement in gaze control, dynamic balance control, and dizziness than a typical outpatient physical therapy regimen.

A randomized, controlled, single-blind pilot trial will be conducted (University of Manitoba Human Research Ethics Board, reference number: H2014:149).

Recruitment

Recruitment and screening (including diagnostics) will be coordinated by physicians and physical therapists in an outpatient center for vestibular rehabilitation. Based on previous pilot research [18], we expect to be able to recruit 56 participants in 12-16 months. Our power analysis is presented in the following section. The inclusion criteria are (1) age 20-50 years, (2) diagnosis of unilateral peripheral vestibular hypofunction based on a detailed neuro-otological and neuro-orthoptic analysis that includes binocular electrooculography with caloric testing; and (3) possession of a home computer with a Windows or Mac operating system. Exclusion criteria include (1) having a central nervous system disorder (cerebrovascular accidents), multiple sclerosis, epilepsy, migraines), axial injury (fractures of the lower extremities or vertebra, advanced hip/knee osteoarthritis), and limited exercise tolerance.

Randomization, Allocation Concealment, and Blinding

This study is designed as a randomized controlled pilot trial. Fifty-six participants will be randomly assigned to either a normative physical therapy comparator control group or to the experimental game-based exercise group. Interventions will begin within 1 week of the baseline assessment. The postintervention assessment will be conducted within 1 week of the final intervention session. Assessors will be blinded to the group assignments.

Interventions

Current treatment for gaze instability due to loss of vestibular ocular reflex consists of rehabilitation techniques that attempt to improve gaze control by enhancing smooth pursuit, saccadic eye movements, and the optokinetic reflex; and by increasing the contribution of the remaining vestibular sensors [19-23]. For this purpose, active goal-directed activities requiring foveation, such as tracking a small visual target or reading aloud and during head movements are essential. The central nervous system must be engaged in purposeful visual fixation and exposed to graded degrees of retinal image slip (ie, head motion

and object motion) [24,25]. A number of studies have used virtual reality systems with large screen displays or DVDs for optokinetic stimulation [26-28]. These optokinetic applications can provide a controlled, configurable stimulus to help clients gradually adapt to motion stimuli and visual scenes that typically induce symptoms of dizziness or disorientation. Current treatment for balance impairments includes the use of various support surfaces such as a compliant sponge pad [19,29,30]. This procedure is used to increase the magnitude and frequency of body sway in a graded fashion (analogous to use of a treadmill for gait training).

The control group will receive a standardized vestibular exercise program consisting of the Herdman gaze stabilization exercises [21,22]. This program is presently a standard of vestibular care. Participants will attend an outpatient physical therapy clinic once a week for 8 weeks. The program also includes a 2-minute home exercise program prescribed 5 times per week for a 12-week duration.

The experimental group will receive the game-based rehabilitation program delivered at home. Modern concepts of learning and neuro-adaptation have been incorporated using a task-specific approach [31,32]. This approach is similar to constraint-induced movement therapy [33], and treadmill locomotor training [34] in that it is a means of repetitive task practice and functional, goal-directed, and meaningful activities of graded intensity and complexity. Our approach will include visual fixation tasks accompanied by head and target motion, thus exposing the individual to graded degrees of retinal image slip.

Utilizing an inexpensive, commercial, motion-sensing mouse that interacts with any computer game or application, we have developed a computer-based rehabilitation platform with a therapeutic gaming application [18,35,36]. The motion-sensing mouse is a small, wireless input device with inertial sensors. These sensors and software acquire instantaneous angular position, which is then used to control the position and motion of the on-screen cursor in a manner identical to a standard computer mouse. Velcro secures the motion-sensing mouse to a headband. With this simple method, head motion is required while the participant is searching, tracking, and interacting with visual targets. This approach provides a highly flexible hands-free game-based treatment tool to train gaze control in standing position and while walking, thus incorporating graded balance demands as well as passive head motion. The use of a human interface device-compliant motion-sensing mouse allows a variety of therapeutic exercises to be coupled to a wide range of inexpensive commercial computer games. Though initially designed for clinical use, the easy-to-use and inexpensive motion-sensing mouse allows this intervention approach to be extended to home settings [18]. The use of computer games with a head rotation input device provides a simple method of graded gaze exercises consisting of visual-fixation and ocular-following tasks accompanied by head motion. We have tested and categorized more than 40 easily accessible commercial computer games. Thus, a broad range of exercise and computer game combinations are available to provide a graded rehabilitation program that includes

1. Predictable cyclic target movement with progression to random moving targets.

2. Small-amplitude movements, with progression to large-amplitude movements.

3. Slow movements, with progression to fast movements.

4. Large targets, with progression to small targets, thus requiring increased precision and foveation.

5. Solid or structured backgrounds (ie, minimal to strong optokinetic stimulation).

The amount of head motion can also be graded using a standard optical mouse and then by using the head-mounted motion-sensing mouse.

Six to eight computer games will be selected for each participant from a pool of commercial games purchased from Big Fish Games platform. Examples of computer games that will be used include the following:

1. Aquaball and Action Ball: Horizontal, single-axis brick buster with slow to moderate speed, low to moderate movement precision, a small to moderate number of distracters, and simple to complex 2D backgrounds.

2. Brave Piglet: Vertical, single-axis game play with moderate speed, moderate to high movement precision, a small to moderate number of distracters, and simple to complex backgrounds.

3. Butterfly Escape: Horizontal, single-axis matching colors with low to moderate speed, low to moderate movement precision, a small to moderate number of distracters, and simple and moving backgrounds.

4. Jet Jumper: Horizontal, single-axis driving and jumping game play with moderate to fast speed, moderate to high movement precision a medium number of distracters, and a complex and moving background.

5. Feeding Frenzy: Two-axis game play with slow motion element, low to moderate movement precision, a moderate to large number of distracters, and a moving background.

These computer games require rapid visual search, tracking of multiple objects in all directions, and active head rotations greater than 100 degrees/s. For example, many of the typical game play elements require game sprite movements covering half to full screen and head rotations up to 60 degrees. These movements are often rapid, completed in 300-500 ms. This equates to head velocities over 100 degrees/s, with precision. In general, there are four types of game objects, namely, (1) game sprite, which is controlled by head rotation movements; (2) game target objects with which to interact (sometimes more than one at a time); (3) distractor objects, which must be ignored; and (4) objects that attack the game sprite and require special attention. These game objects can be stationary or move in predictable or complex trajectories. The player needs to foveate and track multiple targets in short periods and produce head rotations to reposition game sprite with respect to game targets while also avoiding distractors or attacking objects. Usually a player's gaze will be focused on game objects moving

in a direction perpendicular or opposite to that of the game sprite or head motion. During these times, vestibular-ocular reflex (VOR) compensation will be required to maintain gaze stability. There will also be times when the eyes and head will be moving in the same direction and this will require VOR cancellation.

Each participant assigned to the experimental group will attend 3 45-minute clinical therapy sessions. Participants will receive training regarding the specific exercises and activities and use of the motion-sensing mouse and computer games. Initially the games will be played in sitting position with a standard optical hand mouse to assess the level of dizziness or nausea. The head motion mouse will be introduced in the first session. To start the exercise program, games will be selected with relatively slow target movements and with stationary backgrounds and few distractors. Game speed/amplitude, precision, number of distractors, and optokinetic features will be progressed as tolerated.

Balance training will be incorporated into the second session by playing the games while standing on a compliant sponge pad. A compliant sponge pad is now a commonly used unstable support surface in balance re-training of clients with peripheral and central nervous system disorders or older adults with a history of falls. A compliant sponge cannot completely reciprocate the normal body forces beneath the feet as the client moves. This increases the magnitude and frequency of body sway, thus increasing balance demands.

Based on the initial 3 clinical therapy sessions, a home program will be prescribed, customized to the participants' specific balance abilities and tolerance (dizziness). Participants will be instructed to perform their exercise programs 20 minutes per day, 5 days per week. The study's physical therapist will attend the participant's home to set up the motion-sensing mouse and computer games, and to assess the area for fall prevention. Each participant will be instructed to use a chair to provide support. The physical therapist will email each participant weekly to monitor progress, inquire about difficulties with the computer equipment, answer questions, and progress the exercises as outlined earlier.

Recording and Data Analysis

The following information will be collected at baseline prior to start of the interventions: age, gender, work history, history of disease/injury process, and current medications. The assessor will be blinded to participant assignments. The primary outcome measures of the study include measurement of dynamic visual acuity, balance performance, and DHI. The secondary outcome measures of the study include gaze performance and gait analysis.

Dynamic Visual Acuity

The test will measure the ability to see clearly during head rotations of greater than 100 degrees/s. A standardized Early Treatment Diabetic Retinopathy Study eye chart will be used, and the participants will be seated at a viewing distance of 4 m. Participants will be asked to read the letters on the eye chart, first with their head stationary and then when the head is passively rotated horizontally by the clinician/researcher at 2 Hz with the help of a metronome. The difference in the number of lines that participants are able to read when the head is stationary and when rotated will be used as the measure of dynamic visual acuity. A loss of 0-2 lines will be considered normal, whereas a loss of 3 or more lines will represent a loss of VOR function [37]. Moderate to high inter-rater and test-retest reliability has been demonstrated [38,39].

Balance Performance

The test protocol will consist of the following tasks performed in standing position for 45 seconds, first on a fixed floor surface and then repeated while standing on a compliant sponge surface, with eyes open and eyes closed, as described in Desai et al [40]. A force sensor array pressure-sensing mat (Vista Medical Ltd., Manitoba, Canada) will be used to compute the vertical center of foot pressure position in the anterior-posterior and medial-lateral directions. The total path length of center of foot pressure excursions will be computed and divided by total duration (45 seconds) to obtain average velocity. Increased center of foot pressure excursion and mean velocity has been interpreted as decreased stability [40,41]. A composite score will be computed, consisting of the 4 conditions of the modified Clinical Test of Sensory Interaction and Balance: eyes open and closed on fixed and sponge surfaces [29,36,42].

Dhi

The DHI has good test-retest reliability as well as face validity and internal consistency [43].

Gaze Performance

A computerized head-tracking task has been developed for testing gaze performance in standing position on fixed and sponge surfaces and during treadmill walking. Participants will be positioned on a treadmill 100 cm from an 80-cm monitor connected to a computer running the visual tracking application. For a full description and set up, see Szturm et al [18,36]. The head-tracking task involves tracking a bright visual target that moves horizontally or vertically on a computer display in a sinusoidal fashion for several cycles. Two cursors of different colors appear on the monitor. One is the reference or target cursor, which moves at a predetermined frequency of 0.4 Hz and an amplitude of 80% of the monitor width. The second cursor is user controlled and is synced with head rotation via the head-mounted motion-sensing mouse. The task requires 60 degrees of head rotation; average velocity of 50 degree/s and peak velocity of 90 degrees/s. The goal of the visual tracking task is to overlap the 2 cursors as the target cursor moves from left to right or top to bottom of the computer display. In this task, foveation is necessary for the participant to determine the amount of overlap (error) between the moving target and head cursors, and to detect when the target cursor changes direction. The computer application also generates a logged game file to synchronously record coordinates of the target cursor and head rotation at a sampling rate of 80 Hz for offline analysis of head-tracking performance described herein. The head-tracking task will be performed for 45-60 seconds under the following conditions: (1) standing on the fixed floor surface, (2) standing on a sponge surface, and (3) during treadmill walking at a speed 0.9 m/s. Before beginning the walking plus visual-tracking test, participants will walk for 5 minutes for treadmill acclimation,

and to obtain baseline data of a walk-alone condition. During testing a physical therapist will stand behind each participant, ready to provide support if needed. During the treadmill walking test, the participants will wear a safety harness secured from above to a body weight support apparatus (Biodex, model, 945-482, Biodex Medical Systems, New York, USA). Body weight will not be unloaded during testing. The sponge surface and treadmill walking tasks are included to introduce varying degrees of passive head motion [44-46]. Average angular head velocities for a fixed surface has been found to be 3 degrees/s: this increases to 8 degrees/s when standing on the sponge surface and to 30 degrees/s during treadmill walking [18]. When tested on the fixed surface with minimal passive head motion, the performance of the head-tracking task would represent the function of the smooth pursuit system and also requires VOR cancellation (ie, eyes and head moving in the same direction as the target object). When passive head motion is introduced into the visual-tracking task (especially during treadmill walking), VOR compensation is also required to maintain gaze stability [47].

The coordinate data of the computer target motion and the user head rotation (motion-sensing mouse) will be used to compute gaze performance for each head-tracking task. For a full description of the data analysis methods, see Szturm et al [18,36]. A nonlinear least squares algorithm will be used to obtain a sine-wave function of the reference target cursor waveform. The first two cycles of the tracking tasks (45 seconds) will be excluded to allow participants time to acquire the moving target and begin tracking. Head rotation trajectories will be fitted to the sine-wave function, and the coefficient of determination will be computed based on the total and average residual difference between trajectories of the reference and head cursor motions. A value approaching 1 equates to perfect overlap of the two cursors, and excellent gaze performance. The coordinate data of each head-tracking trial will then be processed using custom analysis routines written in MATLAB (version 2010a; MathWorks, Natick, MA, USA).

Gait Analysis

A gait analysis will be conducted to examine whether the exercise program transfers to improvements in walking function. A treadmill instrumented with a pressure mat (Vista Medical, CA, USA) will be used to record vertical foot forces for each step during walking trials of 1 minute at 0.9 m/s, and thus include data for 30 consecutive steps [35,48]. The following spatiotemporal gait variables (average and coefficient of variation through 30 consecutive steps) will be computed: step and swing durations, single support times, step width, and step length.

Participants will be asked to complete daily exercise logs. The study therapist will contact each participant through phone or email on a weekly basis to obtain the exercise logs.

Interventions will begin within 1 week of the baseline assessment (primary and secondary outcome measures). The postintervention assessment will be conducted within 1 week of the final intervention session.

Participants in the experimental group will be given the visual tracking game application for home use. The game software automatically logs reference and head cursor coordinates (gaze performance) during the visual tracking task and saves the data to a coded and time-stamped computer file. Participants will be asked to play the tracking task in standing position at the beginning of every training session. This will involve 3 short tests of 45 seconds each at 3 tracking speeds. The clients can either email the data files to the investigator, or save them on a provided flash drive, which should be returned at a follow-up visit. This will allow us to perform a within individual trend analysis of up to 48 repeated measures for the experimental group.

Statistical Analysis and Power Assessment

Descriptive statistics, including means, standard deviations, frequencies, and percentages, will be used to describe the experimental and control groups on the baseline demographic variables to achieve our first objective, which is to conduct a power analysis of the required sample size to test the difference between the experimental and controls groups at the postintervention measurement. We will test the difference between experimental and control groups on continuously and normally distributed outcome measures using analysis of covariance, with the dependent variable being the postintervention measurement of the outcome, the covariate being the preintervention measurement, and group membership being the between-participant effect. Descriptive statistics, including measures of skewness and kurtosis, will be used to assess departures from the assumptions of a normal distribution of responses. If the distribution contains extreme observations, a robust ANCOVA statistic will be adopted [49]. To test differences between experimental and control groups on discrete outcomes, such as number of times participants feel dizzy during their daily activities, we will use a Poisson regression model, with the dependent variable being the number of relevant events, the covariate being the preintervention measurement, and group membership being the between-participant effect.

To analyze the longitudinal data associated with the second objective, we will use a regression model with time as the random factor [50]. This model will be adopted because it accounts for clustering of repeated measurements within study participants and uses all available longitudinal data (ie, it does not result in case-wise deletion of study participants due to missing observations). Prior to applying this model to the data, we will conduct descriptive analyses of the proportion and pattern of missing data. To assess model fit, we will use penalized measures of the log of the likelihood function, including the Akaike Information Criterion and Bayesian-Schwarz Information Criterion. Initially we will fit a model with only a random intercept for time; subsequently, we will also consider a random slope. The intraclass correlation and the proportion of variation explained by the random effects will be calculated.

The pilot data showed a standardized effect size of .80. Assuming the number of model covariates to be 3, the proportion of variance explained by these covariates to be 10%, and a two-tailed test of the null hypothesis of no group difference at

alpha=.05, we calculated that a sample size of 46 is required. Given an expected attrition rate of 20% over the study observation period for objective 2, we propose to recruit a total of 56 individuals to participate, with equal numbers for the treatment and control groups.

Results

Enrollment for this study was started in August 2014. To date, 36 participants have been enrolled. There have been 6 drop-outs. It is expected that the study will be completed by January 2016 and the first results are expected to be submitted for publication in spring of 2016.

Discussion

The experimental exercise program to be evaluated in this study allows different gaze, head movement, and balance exercises to be coupled with a wide range of commercial computer games. Motivation to perform tedious home programs may be improved with engaging computer games. Our platform is designed to provide client-centered and engaging programs of rehabilitation, and lead to the progression from supervised to unsupervised (monitored) home programs. This study's findings will provide results of effectiveness and permit assessment of the potential for successful home implementation with appreciation of adverse events.

A successful application of this rehabilitation program would help streamline rehabilitation services, leverage therapist time spent with clients, and permit regular practice times at the client's convenience. This research will address a specific, client-centered e-Health application aimed to empower individuals to manage chronic conditions and permit timely detection and intervention tools for monitoring individual and population health. The program may further assist with development of integrated solutions to support a continuum of individual and population-based care and improve accountability and access to long-term rehabilitation services.

Limitations

With few exceptions, commercial computer games do not provide the ability to record the time played, the intensity level, or the player performance, and therefore cannot monitor the actual treatment effort. Thus, compliance will be assessed using participant self-report through completion of exercise logs. While this is common practice, the ability to have games automatically log and send actual time played, scores, and game levels achieved would assist with clinician assessment and progression of the home program.

Gaze performance is based on examination of head movements with respect to object movement. However, eye movements are not recorded and gaze position is not computed. These eye movements are required to determine the contributions of increased VOR gain and adaptations of smooth pursuit and saccadic eye movements. This study is a pre/postcase series design, and no comparisons can be made to existing vestibular rehabilitation programs.

Conclusion

A successful application of this program would help streamline rehabilitation services, leverage therapist time spent with clients and permit regular practice times at the client's convenience. This research will address a specific, client-centered eHealth application aimed at empowering individuals to manage chronic conditions and permit timely detection and intervention tools for monitoring individual and population health.

Conflicts of Interest

None declared.

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Abbreviations

DHI: Dizziness Handicap Inventory
PVD: peripheral vestibular dysfunction
VOR: vestibular-ocular reflex

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Original Paper

Effectiveness of a Mobile Phone App for Adults That Uses Physical Activity as a Tool to Manage Cigarette Craving After Smoking Cessation: A Study Protocol for a Randomized Controlled Trial

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Abstract

Background: Results from studies on the effects of exercise on smoking-related variables have provided strong evidence that physical activity acutely reduces cigarette cravings. Mobile technology may provide some valuable tools to move from explanatory randomized controlled trials to pragmatic randomized controlled trials by testing the acute effectiveness of exercise on quitters under real-life conditions. An mHealth app was developed to be used as a support tool for quitters to manage their cigarette cravings.

Objective: The primary aim of this paper is to present the protocol of a study examining the effectiveness of the Physical over smoking app (Ph.o.S) by comparing the point prevalence abstinence rate of a group of users to a comparator group during a 6-month follow-up period.

Methods: After initial Web-based screening, eligible participants are recruited to attend a smoking cessation program for 3 weeks to set a quit smoking date. Fifty participants who succeed in quitting will be randomly allocated to the comparator and experimental groups. Both groups will separately have 1 more counseling session on how to manage cravings. In this fourth session, the only difference in treatment between the groups is that the experimental group will have an extra 10-15 minutes of guidance on how to use the fully automated Ph.o.S app to manage cravings during the follow-up period. Data will be collected at baseline, as well as before and after the quit day, and follow-up Web-based measures will be collected for a period of 6 months. The primary efficacy outcome is the 7-day point prevalence abstinence rate, and secondary efficacy outcomes are number of relapses and cravings, self-efficacy of being aware of craving experience, self-efficacy in managing cravings, and power of control in managing cravings.

Results: Recruitment for this project commenced in December 2014, and proceeded until May 2015. Follow-up data collection has commenced and will be completed by the end of December 2015.

Conclusions: If the Ph.o.S app is shown to be effective, the study will provide evidence for the use of the app as a support tool for people who are trying to manage cravings during smoking cessation programs. It is anticipated that the results of the study will provide knowledge of how physical activity affects cigarette craving in real-life situations and inform the development and delivery of relapse prevention in smoking cessation treatment.

Trial Registration: International Standard Randomized Controlled Trial Number (ISRCTN): ISRCTN55259451; http://www.controlled-trials.com/ISRCTN55259451 (Archived by WebCite at http://www.webcitation.org/6cKF2mzEI)

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KEYWORDS

behavior change; mHealth app; physical activity; randomized controlled trial; relapse prevention; smoking

Introduction

Although smoking cessation treatments such as behavioral support, nicotine replacement therapy (NRT), and pharmacological treatment have been effective in the short term, the long-term relapse rates are high. The percentages of relapse are between 95% for the cold turkey method and 75% for combined therapies of counseling and pharmacotherapy [1-3]. A systematic review of the effectiveness of smoking relapse prevention [4] indicates that the most effective long-term methods in preventing relapses are self-help materials. Different types of pharmacotherapy showed varied results from short-term to long-term effectiveness and behavioral interventions for relapse prevention appeared effective in the short-term only. The skills approach to behavioral intervention has been widely studied as a method with promising results [5]. Nevertheless, there is no strong evidence to support any specific behavioral intervention as being most helpful to avoid relapses after quitting smoking and there is little research available regarding other behavioral approaches [6]. The latest efforts to develop a taxonomy of behavior change techniques, used in interventions in general [7] and more specificly in smoking cessation interventions [8-10], are expected to help develop stronger evidence in future research in this field.

Exercise has recently been incorporated as a smoking cessation aid into existing programs with promising results. In general, studies examining the effects of exercise on variables related to smoking cessation have focused on either the long-term effects after long interventions or on the acute effects of exercise on smoking-related variables. A recent review [11] of exercise interventions for smoking cessation revealed mixed results. The heterogeneity of the studies reviewed did not allow credible conclusions to be drawn. They focused more on studies that provided data for long-term effects (ie, follow-up data collection at more than 6 months). Within that focus, they identified 15 trials, 3 of which showed significantly higher abstinence rates in a physically active group versus a control group at the end of treatment [12-14]. One of the studies also showed a significant benefit for exercise versus control on abstinence at the 3-month follow-up and a benefit for exercise at the 12-month follow-up [13].

The main reason for relapsing is the craving that smokers experience. Results on the effects of exercise on smoking-related variables have provided strong evidence that physical activity acutely reduces cigarette craving [15-17]. Fourteen reviewed studies that compared a bout of exercise with a passive condition or compared 2 intensities of exercise concluded that even relatively small doses of exercise should be recommended as an aid to managing cigarette cravings and withdrawal symptoms [15]. The results of 12 reviewed studies were similar, showing that cigarette craving was reduced following exercise with a wide range of intensities, from isometric exercise and yoga to activities with heart rates as high as 80%-85% [16]. Using individual participants' data from 17 studies, participants' engagement in physical activity was compared to that of control group participants. The results showed that the effects of all primary studies were in the same direction, with physical activity groups showing a greater reduction in cravings compared with controls, implying strong evidence that physical activity acutely reduces cigarette craving [17].

Evidence of the positive effects of exercise on acute reduction of cravings from experimental laboratory-based studies is strong [15-18], but findings are limited by a lack of long follow-up and artificial settings. Therefore, there is a need to start experimenting with interventions in real-life situations. One effort in this direction is a recent study that used a lab-based scenario with high ecological validity to show that an acute bout of exercise can reduce cravings following concurrent stressors [19]. Similar attempts to collect data from real life for smoking and exercise behavior using ecological momentary assessment methods (EMAs) have also been observed in the recent literature [20-21]. However, the investigation of the effects of short bouts of exercise as a means to acutely manage cravings in real life and the long-term effects of this method on relapse rates has not yet been the focus of any study. Our approach is similar to EMA methods in that it offers real-time data, but it differs in that entries are self-initiated and in how the app also acts as an intervention by offering suggestions to overcome craving and supportive motivation messages. The growing use of mobile technology may provide some valuable tools to move from explanatory randomized control trials that test the acute efficacy of exercise on abstinent smokers under highly controlled conditions to pragmatic randomized control trials that test the acute effectiveness of exercise on quitters under real-life conditions.

Mobile technologies are a means for providing individual-level support to health care users and a promising platform for health interventions [22]. Behavioral intervention technologies offer promising opportunities to expand psychological practice [23]. Several mobile health interventions have been designed to increase healthy behavior (eg, to promote smoking cessation or increase activity levels) and are also cost effective [24]. A review of mobile phone-based interventions for smoking cessation revealed only 4 trials: 2 using SMS text messages and 2 using the Internet and mobile phones. The trials were heterogeneous. However, when the data from both programs were pooled, they found statistically significant increases in both short- and long-term self-reported quitting [25]. A more recent systematic review [26] concluded that SMS-based smoking cessation interventions more than doubled

biochemically verified smoking cessation at 6 months. Moreover, the self-help methods have been indicated in previous literature as effective for long-term effects [4] and the use of mobile phone applications as a self-help method sounds promising.

Therefore, a mobile phone application has been developed for the needs of this study. The aim of the app is to support quitters in managing their cigarette cravings and abstaining over the long term by using exercise as the main behavioral substitution strategy. The mHealth app called "Physical over Smoking (Ph.o.S)" includes a data collection mechanism to collect real-time data regarding relapses. The Ph.o.S app was developed based on evidence-based practices for relapse prevention after smoking cessation. Following that, a trial has been designed to test the effectiveness of the mHealth app.

Trial Aim

The aim of this study is to present the protocol of a study that assesses the effectiveness of the Ph.o.S app, which helps abstaining users to manage their cigarette cravings. The study will examine the quit rates of abstaining users of the app versus the quit rates of a comparator group for a 6-month period after quitting smoking. The trial's main hypothesis is that users of the Ph.o.S app will have higher 7-day point prevalence abstinence rates during the follow-up measures in comparison to the comparator group (trial registration number ISRCTN55259451).

Four additional hypotheses have been proposed:

1. Users of the Ph.o.S app will have fewer self-reported relapses during the follow-up measures than the comparator group will.
2. Users of the Ph.o.S app will report higher self-efficacy from being aware of experiencing cravings than the comparator group will.
3. Users of the Ph.o.S app will report higher self-efficacy in managing cravings in relapse situations than the comparator group will.
4. Users of the Ph.o.S app will report a higher power of control to manage cravings in everyday situations than the comparator group will.

Trial Design

This study is a 2-armed randomized clinical trial. Participants identified as eligible and who give their consent to participate will receive a smoking cessation program consisting of 3 group sessions (once per week). After the quit day, participants will be randomly assigned to 2 groups. Both groups will have a separate fourth session where they will receive training on relapse prevention and a plan to cope with cravings. The intervention group will receive an additional short training session on how to use the Ph.o.S app as an additional support tool whenever they experience cravings in their everyday life during the follow-up period. Preintervention, postintervention, and 6-month follow-up assessments will be conducted. Table 1 displays an overview of the study's timeline and the schedule of enrollment, interventions, and assessments. Multimedia Appendix 1 displays the detailed measurement time points for each group.

Table 1. Study timeline: overview of schedule of enrollment, interventions, and assessments.

Timeline points	Schedule				
	Screening for eligibility	Intervention: quit smoking (3 sessions)	Quit day: allocation	Intervention: manage cravings (4th session)	Follow-up assessment
$-t_3$	X				
$-t_2$		X			
$-t_1$		X			
t_0			X		
t_1				X	
t_2 (3 days after)					X
t_3 (1 weeks after)					X
t_4 (2 weeks after)					X
t_5 (3 weeks after)					X
t_6 (4 weeks after)					X
t_7 (12 weeks after)					X
t_8 (24 weeks after)					X

Methods

Study Setting

The study setting is the Jyväskylä Community Primary Health Care Center in Central Finland.

Eligibility Criteria

All interested participants are screened for eligibility by completing a short battery of questions online. Noneligible participants are advised to contact their doctor or nurse for help. Participants are adults (between the ages of 18 and 65) who have been smokers for at least 1 year and who smoke more than 10 cigarettes per day. (Those who use snus only are excluded.) They should have no other addictions (ie, alcohol, prescription drugs, or illegal drugs) according to the behavioral screening tool NIDA Quick Screen V1.0 [27]. Eligible participants should be addicted to nicotine, with a score of more than 4 out of 10 on the Tobacco Dependence Screener (TDS) [28]. TDS is a 10-item self-report questionnaire, and it generates a continuous dependence score. It demonstrates acceptable reliability in different samples as well as validity because it has a high association with smoking heaviness measures (eg, number of cigarettes smoked per day, carbon monoxide levels) and years of smoking [28-29]. Participants should have a strong motivation to quit and score more than 3 out of 7 on the Motivation to Stop Smoking Scale (MTSS) [30]. The MTSS is a single-item scale assessing an individual's motivation to quit smoking, combining aspects of desire and intention to quit. Scores range from 1 (I don't want to stop smoking) to 7 (I really want to stop smoking and intend to in the next month). Participants are also screened for active psychological distress by completing the 12-item General Health Questionnaire [31] and those scoring more than 20 out of 36 are excluded [32,33] because they have lower quit rates [34,35]. Participants are screened for health risks if they increase their physical activity by completing the Physical Activity Readiness Questionnaire [36]. (Those answering yes to any question will be excluded.) Nevertheless, participants who were able to provide permission to exercise from their doctor were accepted to the program. For example, if their blood pressure, heart condition, or bone or joint problem is well controlled with drugs and their doctor says it is safe to exercise.

Finally, the Gold Standard Monitoring Form used by the National Health Service in the United Kingdom is completed for each participant [37]. The form records useful information regarding participants' personal details, information about the type of intervention, any pharmacological support (eg, NRT, varenicline), use of other nicotine products (eg, snus), and the outcome. Participants currently trying to quit or taking a smoking cessation medication (eg, NRT, wellbutrin, or varenicline) or receiving any other form of treatment (eg, Web-based, app-based, or other) are not excluded as long as they agree to follow our methods.

Recruitment, Randomization, and Allocation

Recruitment will occur through referrals from the health care units in the Jyväskylä area as well as via the Web pages of Central Finland Respiratory Association, the City of Jyväskylä, Radio Jyväskylä, and Yle News. All interested participants will be screened for eligibility. The enrollment period ended in May 2015. The first 50 eligible participants will start the smoking cessation intervention, which consists of 3 weekly counseling sessions and will help them set a quit date. The rest of the eligible participants will be put on a waiting list and invited to participate in case of dropouts before the quit day/randomization time point. The first 50 participants to reach the quit day/randomization time point will then form the intention-to-treat group that will continue in the study. After the quit day/randomization, participants will be assigned to an experimental group (25 participants) and a comparator group (25 participants), and both groups will separately have 1 more counseling session on how to manage cravings. In this fourth session, the only difference in treatment between the groups is that the experimental group will have an extra 10-15 minutes of guidance on how to use the Ph.o.S app to manage cravings during the follow-up period. Both groups will be followed up for the main and secondary efficacy outcomes of interest for 6 months. The method of randomization is a set of numbers generated by online software [38]. The principal investigator will generate the allocation sequence and the study nurse will assign participants to the groups. The study nurse is blind to the allocation sequence until the third session and the principal investigator is blind to the participants and their data up to the first follow-up measure.

Interventions

Smoking Cessation and Relapse Prevention Interventions

The presentation of the smoking cessation intervention content has been based mainly on the taxonomy of behavior change techniques in interventions [7] and the taxonomy of behavior change techniques used as support for smoking cessation [39]. The theoretical frameworks that the techniques are based on are control theory [40], the social-cognitive theory [41], the theory of planned behavior [42], and the motivational interviewing technique [43]. During the first session, baseline data are collected and then a group or individual motivational interview session takes place for about an hour, aiming to motivate participants to take further responsibility for making important health-related behavior changes (ie, to gradually decrease smoking and be more physically active). During the second session, participants discuss the barriers for behavior change and they brainstorm facilitators to apply. In the third session they set specific goals and make plans for behavior change. In this meeting, participants also set a quit day and they get training on the use of pedometers. The 3 sessions are scheduled weekly. The intervention is a shorter version of a previously effective intervention named "No more smoking! It's time for physical activity" [44-45]. Multimedia Appendix 2 contains a more detailed overview of the contents of the intervention used to help participants quit smoking.

All sessions are delivered by the study nurse, who has previous experience with counseling smokers to quit. Multimedia Appendix 3 presents the content of the fourth session that covers relapse prevention, which is held for the participants within 1 week after their quit day. The aim of this fourth session is to help participants develop a cravings management plan to prevent relapses. Similarly, the presentation of the fourth session's

content has been based on the taxonomy of behavior change techniques used as support for smoking cessation [39] and in general interventions [7]. The theoretical frameworks that the techniques are grounded in are the relapse prevention model [46,47], control theory [40], social-cognitive theory [41], and the theory of planned behavior [42].

Database and Data Collection Mechanism of the Ph.o.S App

The mHealth mobile phone application, Ph.o.S, and its data collection mechanism was developed by a software designer affiliated with the University of Jyvaskyla, as a project researcher at Agora Center. Additional support for the development was provided for graphic design and user interface design by 2 students from the IT department. Table 2 presents examples of the database contents by category (eg, introductory messages, physical activities, and motivational messages). The database contents are based on the relapse prevention model [46,47] and the behavior change techniques for smoking cessation via text-messaging intervention [10]. Additionally, the Ph.o.S app was designed according to the principles of persuasive systems design [48]. For example, the principles of tailoring [49], personalization, suggestion, praise, trustworthiness, and expertise have been used in the design of the application. The application database includes a pool of 57 introductory messages, 49 motivational messages, and 64 physical activities, all of which are coded to appear according to the users' profile and status. Introductory and motivational messages were reviewed by 3 experts on health and exercise psychology and physical activities by 2 experts on sport and exercise science. Multimedia Appendix 4 includes examples of screenshots of the sequence from a sample usage session.

During the fourth session, experimental group participants are instructed to use the application whenever they experience cigarette cravings and to give feedback for every use. As a result, data entries are self-initiated whenever participants use the app. The Ph.o.S. app is installed on participants' personal mobile phones, but if they do not have a mobile phone, then a device is provided by the study team. No incentives are provided to use the app or to complete data during the entire follow-up assessment period. When they need to, participants can use the app without an Internet connection. All information is uploaded to the server as soon as the device is connected. Participants' identification number and profile settings are entered when the Ph.o.S app is installed on their mobile phones and used for the first time. Profile settings include gender, age, weight, and height, days since quit date, the origin of the decision to quit, and heavy and moderate intensity physical activity history during a typical week (screenshot 1, Multimedia Appendix 4). After that, every time a user experiences a craving situation, specific information regarding mood (eg, positive, neutral, negative), place (eg, home, at work, outdoors), and social environment (eg, alone, not alone) are requested (screenshot 2, Multimedia Appendix 4). According to users' profile information and the situational status, a variety of animated physical activities, matched with introductory and motivational messages, are suggested. Physical activities were animated to model/demonstrate the suggested activities in a visual form (screenshot 3, see Multimedia Appendix 4). Each time the participant uses the app, he or she is asked to provide feedback on the effectiveness of the app to manage the craving. Answer options are the following: "Yes it worked," "No, but I'll manage," or "No, I relapsed" (screenshot 4, see Multimedia Appendix 4). Multimedia Appendix 5 shows the CONSORT checklist for this study.

Table 2. Ph.o.S app: examples of database content.

Category	Examples	Behavior change techniques
Introductory messages	Researchers have found 70 poisonous chemicals in cigarettes which cause cancer. Stay healthy!	Provide information on consequences of behavior in general
	Your shortness of breath when using stairs is much less now!	Provide information on consequences of behavior to the individual
	Ex-smokers have less than two cravings a day. You can manage this craving!	Provide normative information about others' behavior
	You are a good example to others every day you stay away from smoking!	Prompt identification as role model/position advocate
	Is it worth giving up what you've worked so hard for? Definitely not!	Prompt anticipated regret
	Walk proudly! You are doing an amazing job quitting!	Prompt rewards contingent on effort or progress toward behavior
Physical activities	Walking: Walk and every 10 steps say: You can make it!	Behavioral substitution + Prompt self-talk
	Brisk walking: Walk briskly and every 15 steps say: You can do this!	Behavioral substitution + Prompt self-talk
	Stairs: Find a stairway to go up and down!	Behavioral substitution
	Breathing: Inhale for a count of 4, then exhale for a count of 4.	Behavioral substitution
	Tension release: Grab a ball and squeeze it with your right hand and then with your left hand.	Behavioral substitution
	Stretching: Stretch your upper arms. Hold for 10 seconds.	Behavioral substitution
	Balance: Balance on your right leg and then on your left leg for 5 seconds.	Behavioral substitution
	Strength: Do as many push-ups as you can in a row.	Behavioral substitution
	Isometric: Hold the superman position for 5 seconds.	Behavioral substitution
	Dance: Listen to your favorite song and dance!	Behavioral substitution
	Gardening, cleaning home physical activities: Do some housework!	Behavioral substitution
Motivational messages	Pain is temporary. Quitting is forever!	Facilitate relapse prevention and coping
	I can keep going!	Enhance self-regulation
	Never be a slave to cigarettes again!	Motivation to remain abstinent
	Do not go back! You are a permanent ex-smoker now!/You are a healthy ex-smoker!	Motivation to remain abstinent
	Concentrate on your goal!	Maintain engagement
	It's not the situation; it's your reaction to the situation!	Provide information on withdrawal symptoms
	I want to feel like a winner, not miserable after 3 minutes!	Prompt anticipated regret
	My actions are always within my control.	Self-regulation/self-control

Participant Retention

To prevent missing data, an effort will be made to retain the participants in the trial for the follow-up data collection. However, participants who withdraw their consent will discontinue their participation in the study. Moreover, participants who attend less than 2 out of 3 sessions of the quit smoking intervention will be excluded from the final analysis. Participants who drop out of the trial or who are lost for follow-up are considered to be smoking. The flow of the participants is presented in Figure 1.

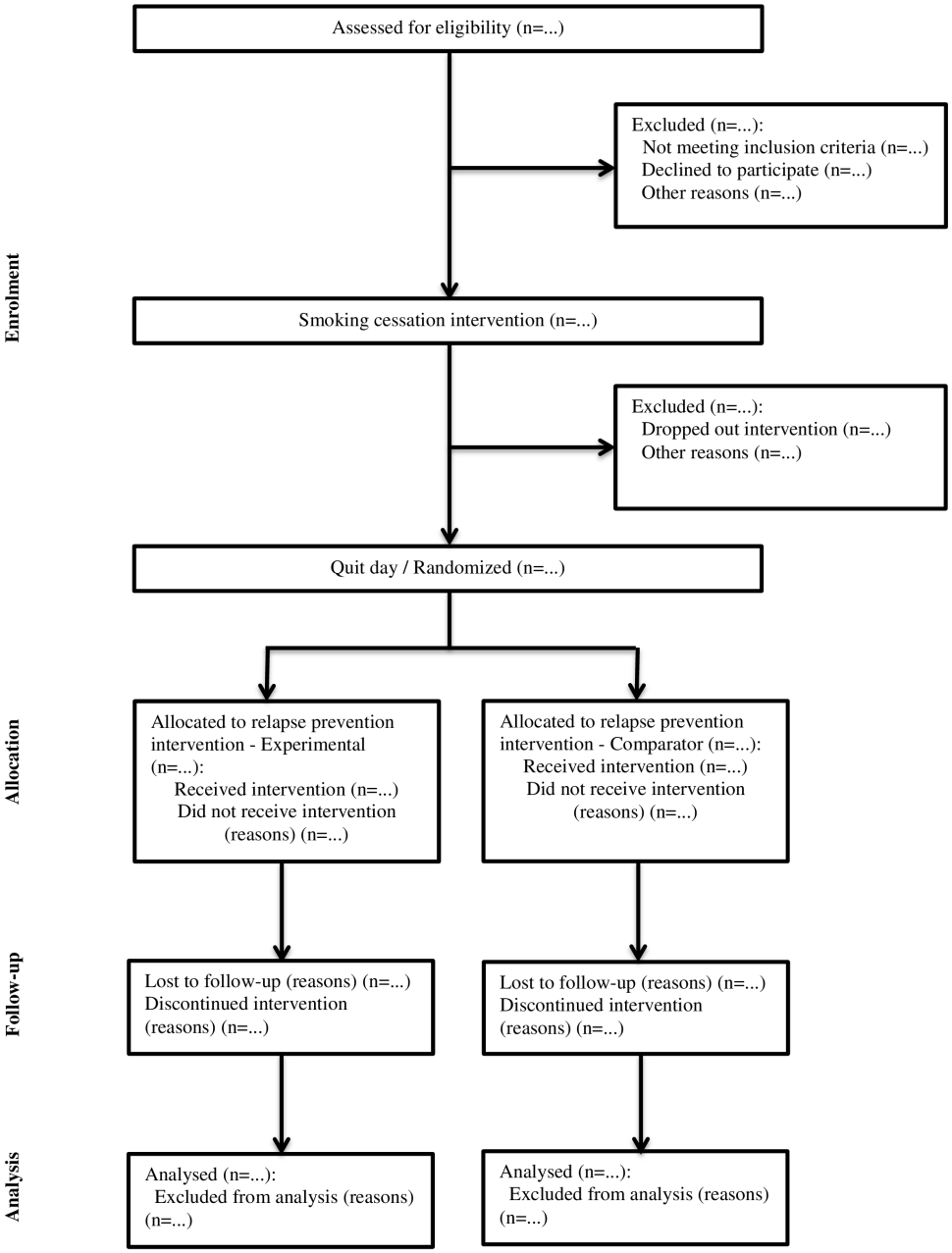
The strategies for monitoring and improving adherence to intervention protocols include the following:

1. Face-to-face adherence reminder. Study participants will be well informed during the first session about the expectations regarding study procedures before they sign the informed consent form. Moreover, a reminder about

the expected participation during the follow-up measures takes place during the fourth session when they are asked about their preferred way of follow-up data collection (eg, telephone, mail, email or SMS).

2. Providing feedback about how well subjects adhere to protocol and achieve target goals whenever possible. During the follow-up period a short SMS or email message will be sent to all participants (ie, both experimental and comparator). The follow-up messages start with an acknowledgment of the participant's contribution to date (ie, how well they adhere to the protocol), followed by a motivational message that is individualized depending on their progress (ie, relapse or no relapse). Next, information on how to fill out the follow-up questionnaire is provided and, finally, the date of the next follow-up point is reported.
3. Monitoring adherence to the Ph.o.S app use and intervening when adherence problems emerge. In the experimental group, adherence is monitored by recording through the data collection mechanism of the Ph.o.S app. When participants do not use the Ph.o.S app for 1 week, they are asked, via SMS or email, to verify if they use the Ph.o.S app, to check if there is a technical problem, or if they have stopped using the Ph.o.S app. If they answer that they do not use the app, then they are sent a link to a short questionnaire in order to assess the reasons for discontinuing use of the app. Participants are considered dropouts when they do not respond to the invitation emails for measurement but not when they stop using the app.

Figure 1. Study flowchart: The diagram illustrates the flow of participants.



Results

Primary Efficacy Measure

Point prevalence abstinence (PPA) will be the primary outcome and the primary efficacy parameter for the study. The main outcome analyses are based on 7-day PPA (ie, reported abstinence of at least 7 days prior to each scheduled follow-up). PPA and prolonged abstinence are closely related and can be interconverted with moderate accuracy [50]. Self-reports of smoking behavior over the last 7 days are collected from participants at the fourth session (t_1), 3 days after the fourth session day (t_2), weekly after the fourth session day through week 4 (t_3-t_6), and at weeks 12 (t_7) and 24 (t_8) after the fourth session day. Self-reported PPA at the t_1 and t_8 follow-ups are verified by saliva cotinine (with a cutoff value of 10 ng/mL) for stated abstinence using NicAlert saliva-cotinine tests [51]. The timeline follow-back for smoking is used to assess daily smoking intensity [52] in a small 1-week calendar form on paper to record how many cigarettes they smoked each day in the last 7 days.

Secondary Efficacy Measures

The secondary efficacy parameters for the study include the following measures:

1. The self-reported number of relapses, which is assessed through a single item asking participants to complete the number of relapses they had the last 7 days. Data are collected from participants at the fourth session (t_1), 3 days after t_1 (t_2), weekly from t_1 through week 4 (t_3-t_6), and at weeks 12 (t_7) and 24 (t_8) after t_1.
2. The self-reported number of cravings, which is assessed through a single item asking participants to complete the number of cravings they experienced in the last 7 days. Data are collected from participants at the fourth session (t_1), 3 days after t_1 (t_2), weekly from t_1 through week 4 (t_3-t_6), and at weeks 12 (t_7) and 24 (t_8) after t_1.
3. Self-efficacy on being aware of experienced cravings, which is assessed with the single item: "How well are you aware of your cigarette cravings?" Answers are given on a 10-point scale from 1 (very poorly) to 10 (very well). Data are collected from participants before the quit day (-t_2), at t_1, t_2, weekly from quit day at t_3-t_6, and at weeks t_7 and t_8.
4. Self-efficacy on managing cravings, which is assessed with the single item: "How well do you manage your cravings?" Answers are given on a 10-point scale from 1 (very poorly) to 10 (very well). Data are collected from participants at -t_2, t_1, t_2, weekly at t_3-t_6, and at weeks t_7 and t_8.
5. Power of control in managing cravings is assessed with 6 items. An example item is "If I am in a situation where I celebrate with my friends...," then, the answer for the experimental group is "It will be more difficult to use Ph.o.S. app to control my craving for tobacco," and for the comparator group is "It will be more difficult to do something to control my craving for tobacco." Answers are given on a 7-point scale from 1 (totally agree) to 7 (totally disagree). The other items present similar tempting situations like when participants are stressed or angry. Data are collected from participants before the quit day at -t_2 and -t_1 and after t_1 at weeks t_2, t_6, t_7, and t_8.

Additional Measures

Additional measures of mainly psychological variables will be collected in order to use them as possible explanatory variables of the primary or secondary efficacy measures. Moreover, usability, fidelity, and data extracted from the phone database will be collected for validation purposes.

1. Physical activity behavior. Self-reported data regarding participants' current physical activity behavior as recorded with the International Physical Ativity Questionaire (IPAQ) [53] will be collected at -t_2 and -t_1 and at weeks t_7 and t_8. Self-reported physical activity behavior through self-reports, before the quit day at -t_2 and -t_1, will be verified by measuring their step counts (via pedometer Omron HJ-152) the last week before measurement day. IPAQ records the physical activity of 4 intensity levels for the last 7 days: (1) vigorous-intensity activities such as aerobics, (2) moderate-intensity activities such as leisure cycling, (3) walking, and (4) sitting.
2. Relapse situation efficacy as recorded with the Relapse Situation Efficacy Questionnaire (RSEQ) [54] will be assessed before the quit day at -t_2 and -t_1. RSEQ consists of 75 simple items assessing participants' confidence in their ability to resist the temptation to smoke in a wide variety of contexts. A 4-point response scale is used from 1(not at all confident) to 4 (extremely confident).
3. Attitude toward, intention to, and perceived behavioral control of increasing physical activity behavior will be measured through a self-reported questionnaire before and after quit day at -t_2 and t_1. Similarly, attitude toward, intention to, and perceived behavioral control of quitting smoking will be measured before quit day at -t_2. Likewise, attitude toward, intention to, and perceived behavioral control of craving management will be measured during the fourth session at t_1. Attitude toward all 3 behaviors (ie, increasing physical activity, quitting smoking, and managing cravings) is assessed with 6 items using a 7-point semantic differential scale (ranging from -3 to +3): ''To (behavior) in the following month will for me be...'': good-bad, unpleasant-pleasant, wise-silly, easy-difficult, healthy-unhealthy, important-not important. Similarly, intention is measured with 3 items: "In the following month...": (1) "I intend to (behavior)," (2) "I will try to (behavior)," (3) "I plan to (behavior)." Answers are given on a 7-point scale ranging from 1 (very likely) to 7 (very unlikely). Correspondingly, perceived behavioral control is measured with 4 items, for example: "For me to (behavior) in the next month is...," and rated on a 7-point scale, with participants answering from 1 (easy) to 7 (difficult).
4. Usability of the Ph.o.S app for the experimental group only will be collected through the System Usability Scale (SUS) self-reported questionnaire [55] 1 week (t_3) and 1 month (t_6) after participants have started using it. SUS is a 10-item

scale giving a global view of subjective assessments of usability. Answers are given on a 5-point scale from 1 (strongly agree) to 5 (strongly disagree). Moreover, a follow-up qualitative assessment when the user stops using the app is performed in order to determine the reasons for discontinuation and possible other means they use to manage cravings effectively.

5. All participants are asked fidelity check questions during the follow-up measure at t_2, t_3, t_6, t_7, and t_8. Questions are related to additional help (eg, pharmacological, mHealth apps) used to manage cigarette cravings. Additional questions for the experimental group are related to the ways they use the Ph.o.S app.
6. Information from the Ph.o.S app is also collected constantly during the follow-up period for the experimental group from the time they start using it (t_2-t_8). Data, such as frequency of use and frequency of successful and unsuccessful efforts to manage craving through the suggested solutions, will be extracted and used to verify the corresponding self-reported questions. Additional data will be extracted for descriptive purposes.

Data Collection Procedure

Screening data at time point -t_3 will be collected via an online questionnaire to determine eligibility for all participants who respond via the various recruitment strategies. The questionnaire consists of a short overview of the aim of the study and questions regarding the person's addictions, quitting history, willingness to quit, general health, and readiness for physical activity. The website used to assess eligible participants is a safe and secure site called Fluid Surveys. People who are eligible, based on the online assessment, receive a follow-up telephone call from the study nurse to verify eligibility and will be invited to come to the health center to complete the consent to participate and the baseline measures.

Baseline data and behavioral and psychological self-reported data at time points -t_2, -t_1, and t_1 will be collected during the first, third, and fourth sessions of the smoking cessation intervention in a paper-and-pencil format, administered by the study nurse in a quiet area of the unit. Follow-up data at all follow-up time points (t_2-t_8) will be collected via online questionnaires after an invitation from the research assistant to participants, via email or SMS, to complete the questionnaires. In addition, data from the phone app users (experimental group only) will be collected during the follow-up period. Users' phone data will be uploaded daily to a secure university server reserved for this purpose.

Once participants have participated in the final meeting, the researchers will make every reasonable effort to follow the participant for the entire follow-up period of 6 months. Participants may, however, withdraw from the study for any reason at any time. However, early discontinuation of Ph.o.S app for any reason is not a reason for exclusion from the study.

Data Management

The University of Jyväskylä is responsible for storing and protecting the research data. The research registry is kept at the University of Jyväskylä in a locked cabinet. All electronic data will be stored on a university computer with password protection. Only the principal investigator and the research assistant will have access to the computer-based data. All participants will be assigned a code number in order to protect their confidentiality. All stored data will be under the unique code number. All data will be entered electronically. Original study forms will be entered and kept on file at the participating site. Participant files are stored in numerical order and in a secure and accessible place and manner. Participant files will be stored for a period of 3 years after completion of the study.

Statistical Methods

Generalized linear mixed models will be used to examine differences between the groups on the dichotomous primary outcome variable: abstinent versus smoker status at the 6-month follow-up time point (ie, t_8). The differences at all follow-up time points will also be examined, but the 6-month follow-up is considered to be the primary focus. Descriptive statistics will be used to report process data. Any covariates identified in the preliminary analyses will be added to the model. Linear mixed models will be used to analyze the secondary efficacy measures for data that are normally distributed or are approximate to a normal distribution (via data transformations). For count variables, a generalized Poisson mixed model will be applied. Alternatively, we will use nonparametric tests by time point separately.

Finally, 3 separate logistic regressions on the primary dichotomous outcome (abstinent-smoker) will be performed using the theory of planned behavior (TPB) constructs of quit smoking behavior and increased physical activity behavior at the -t_2 time point and the TPB constructs of manage cravings behavior at the t_1 time point as predictors. Linear regressions for the continuous variables of the secondary efficacy outcomes will be used to test the predictive ability of all TPB constructs. All analyses will be conducted using intent-to-treat principles with participants remaining in their originally assigned groups after randomization regardless of adherence or protocol deviation. This is done to increase the likelihood that group differences are due to the intervention and unaffected by biases that lead to an overestimation of the intervention effect [56].

Power

We determined the power for our primary hypothesis, which stated that the 7-day PPA at the 6-month follow-up would be higher in the experimental group than in the comparator group. In order to detect a difference of 10% in quit rates between the experimental and comparator groups, with an expected quit rate of 25% in the comparator group, the study would require, at 80% power with a two-sided p value of 0.05, a sample size of 326 participants. Given the budget and time limitations as well as the location (eg, a small city in a sparsely populated area, which makes recruitment a challenge), a sample size of 50 has been set by the research group as a more realistic number when taking into account the available resources. Given the sample size of 50 participants equally assigned to the groups, we calculated the smallest PPA that we could detect with power greater than 0.80. Assuming that the abstinence in the comparator group will be 25% [1-3], power analysis through

online calculations [57] indicate that we will be able to detect a significant difference between the experimental and comparator groups with power greater than 0.80 if the PPA in the experimental group is 59% or higher. The percentage of 59% required to obtain statistical power is considered to be very high. At the end of the study we will re-estimate the power of the final percentage differences between groups in the effectiveness report, depending on the exact final sample size as well. Nevertheless, the sample size is considered as an a priori limitation to the study.

Ethics and Dissemination

Ethics approval has been obtained from the Ethics Committee of the Central Finland Health Care District (Keski-Suomen Sairaanhoitopiirin Eettinen Toimikunta).

Consent

All eligible participants are given a copy of the information sheet and informed consent form to read. The information sheet provides a summary of the research study and the informed consent document states what the individual is about to participate in, the individual's rights as a research participant, and information about confidentiality. The study nurse explains all aspects of the study and answers all of the participant's questions regarding the study. If the person chooses to participate in the study, that person will be asked to sign the informed consent form. No study procedure is performed prior to signing the informed consent form. Subjects who refuse to participate or who withdraw from the study are treated without prejudice. The reason for refusal or withdrawal will be noted on the form if reported.

Confidentiality

All study-related information will be stored securely at the study site under a coded identification number in order to maintain participant confidentiality. All records that contain names or other personal identifiers, such as screening data for eligibility and informed consent forms, will be stored separately from study records identified by code number. All local databases are secured with password-protected access systems. Forms, lists, logbooks, appointment books, and any other listings that link participant identification numbers to other identifying information will be stored in a separate, locked file in an area with limited access. Participants' study information will not be disclosed to third parties.

Discussion

This trial and its findings will contribute to the evidence available to inform the development and delivery of relapse prevention on smoking cessation treatment. The primary objective of the study is to contribute to the literature calling for the development of mHealth applications that support individuals in remaining abstinent after they quit smoking. The overall implementation of the project can have an impact on participants' motivation to initiate and adhere to physical activity. Increasing physical activity can reduce health-risk factors and improve self-esteem as well as quality of life. The use of the Ph.o.S app is not restricted to any specific place, time, situation, or quitting method and it is free to use. It can also contribute to the reduction of health care costs. In addition to the impact on public health, the project will have a significant contribution to future research on how physical activity affects cigarette cravings in real-life situations and will offer the potential to improve the understanding of the mechanisms that underlie these effects. Nevertheless, the relatively small sample size due to time and budget limitations, as well as the recruitment and engagement difficulties that are common with smokers in clinical trials [58], is a threat to the power of the trial.

Trial Status

Recruitment for this project commenced in December 2014 and proceeded until May 2015. Follow-up data collection has commenced and will be completed by the end of December 2015.

Acknowledgments

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Authors' Contributions

MH, MV, TL, TK, and KK conceived of the original idea for the trial and also sought and obtained funding. MH, HT, MV, TL, TK, and RH wrote the study protocol. This protocol paper was written by MH with input from all co-authors. HT manages the day-to-day running of the trial, including all participant follow-ups. RH will undertake all data analyses. All authors read and approved the final manuscript.

Conflicts of Interest

None declared.

Multimedia Appendix 1

Study timeline: Schedule of enrollment, interventions, and assessments.

[DOC File, 27KB - resprot_v4i4e125_app1.doc]

Multimedia Appendix 2

Overview of the smoking cessation interventions content (Sessions 1, 2, and 3).

[DOC File, 18KB - resprot_v4i4e125_app2.doc]

Multimedia Appendix 3

Overview of the relapse prevention intervention content (Session 4).

[DOC File, 19KB - resprot_v4i4e125_app3.doc]

Multimedia Appendix 4

Ph.o.S. flow: Examples of screenshots.

[PNG File, 330KB - resprot_v4i4e125_app4.png]

Multimedia Appendix 5

CONSORT Checklist form.

[PDF File, 1MB - resprot_v4i4e125_app5.pdf]

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Abbreviations

EMA: Ecological Momentary Assessment Methods
IPAQ: International Physical Ativity Questionaire
MTSS: Motivation to Stop Smoking Scale
NRT: nicotine replacement therapy
Ph.o.S: Physical over Smoking app
PPA: point prevalence abstinence
RSEQ: Relapse Situation Efficacy Questionnaire
TDS: Tobacco Dependence Screener

TPB: theory of planned behavior

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Protocol

Mobile Phone Apps for University Students With Hazardous Alcohol Use: Study Protocol for Two Consecutive Randomized Controlled Trials

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Abstract

Background: About 50% of university students overconsume alcohol, and drinking habits in later adulthood are to some extent established during higher educational studies. Several studies have demonstrated that Internet-based interventions have positive effects on drinking habits among university students. Our recent study evaluated two mobile phone apps targeting drinking choices at party occasions via personalized feedback on estimated blood alcohol concentration (eBAC) for students with hazardous drinking. No changes in drinking parameters were found over a seven-week period apart from an increase in number of drinking occasions among men for one of the apps tested. Up to 30% of the study participants drank at potentially harmful levels: higher than the national recommended number of standard drinks per week (a maximum of 9 for women and 14 for men) in Sweden.

Objective: (1) To evaluate improved versions of the two mobile phone apps tested in our prior trial, in a new, 3-armed randomized controlled trial among university students with at least hazardous drinking habits according to the Alcohol Use Disorders Identifications Test (AUDIT; Study 1). (2) After 6 weeks, to target study participants showing alcohol consumption higher than the national recommended levels for standard drinks per week by offering them participation in a second, 2-armed randomized trial evaluating an additional mobile phone app with skill enhancement tasks (Study 2). (3) To follow participants at 6, 12 and 18 weeks after recruitment to Study 1 and at 6 and 12 weeks after recruitment to Study 2.

Methods: Two randomized controlled trials are conducted. Study 1: Students are recruited at four Swedish universities, via direct e-mail and advertisements on Facebook and student union web sites. Those who provide informed consent, have a mobile phone, and show at least hazardous alcohol consumption according to the AUDIT (≥6 for women; ≥8 points for men) are randomized into three groups. Group 1 has access to the Swedish government alcohol monopoly's app, Promillekoll, offering real-time estimated eBAC calculation; Group 2 has access to a Web-based app, PartyPlanner, developed by the research group, offering real-time eBAC calculation with planning and follow-up functions; and Group 3 participants are controls. Follow-up is conducted at 6, 12 and 18 weeks. Study 2. Participants who at the first 6-week follow-up show drinking levels higher than 9 (W) or 14 (M) standard drinks (12 g alcohol) per week, are offered participation in Study 2. Those who consent are randomized to either access to a skills training app, TeleCoach or to a wait-list control group.

Results: Latent Markov models for Study 1 and mixed models analyses for Study 2 will be performed. Study 2 data will be analyzed for publication during the spring of 2016; Study 1 data will be analyzed for publication during the fall of 2016.

Conclusions: If mobile phone interventions for reducing hazardous alcohol use are found to be effective, the prospects for positively influencing substance use-related health among university students can considerably improve.

Trial Registration: ClinicalTrials.gov http://clinicaltrials.gov/ct2/show/NCT02064998 (Archived by WebCite at http://www.webcitation.org/6dy0AlVRP)

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KEYWORDS

randomized controlled trial, universities, alcohol abuse, prevention, mobile phone, eHealth, mHealth

Introduction

Research studies have shown that consumption of alcohol is higher during the years at university than at any other age [1]. Furthermore, it appears that students at college and university establish their future adult alcohol habits [2]. Individual, social and environmental factors affect this development [3]. Several studies have demonstrated that effective intervention approaches, both at individual and group levels, are available to reduce hazardous drinking among students. These methods are well documented internationally [4,5] and as well as nationally in a meta-analysis [6].

A major behavior change component for reducing problematic alcohol use in general is screening with brief intervention (SBI), where the primary goal is to moderate consumption to sensible levels and to eliminate harmful drinking practices in order to reduce negative outcomes of drinking [7]. At minimum, SBI includes only screening of drinking habits, followed by clear and direct feedback. It can also include detailed conversations covering setting up goals, use of behavioral modification techniques, self-help exercises, and continual reinforcement [8]. For university students, effective interventions are termed brief motivational interventions (BMIs), providing personalized feedback on individual drinking habits and their consequences, based on self-monitoring, as well as exploration of motives for using alcohol. Specific behavior change components include the personalized feedback aspect, particularly normative feedback, in relation to students in the same university context; studies of feedback on Blood Alcohol Concentration (BAC) have shown mixed support [9].

New Technology

Despite availability of interventions, only a small percentage of students seek help for problem drinking [10]. College students with heavy episodic drinking (HED) at least once a month have been found to prefer computerized methods [11]. Digital interventions for alcohol problems, regardless of delivery mode, offer small but meaningful effects [12-18]. They are as effective as alternative interventions offered face-to-face by a live counselor when compared with controls but, in direct comparisons, face-to-face interventions have been shown to be more effective [17,18].

Mobile Apps

In recent years, mobile phones have offered constant access to hand held computers. Easy to use mobile phone apps can fill a variety of functions, such as games and other entertainment. Depending on its function, the app may or may not require an Internet connection. Apps have been used for registering weight for obesity control [19] and, in a guided version, to help users with behavioral activation in their own chosen valued direction [20]. Research on mobile phone apps for reducing alcohol consumption is in its infancy. Over 3000 apps on alcohol-related topics are available, but these usually have no therapeutic purpose and even provide incorrect information, for example regarding blood alcohol content [21]. An analysis of user experiences of 87 Android apps showed, however, that these in some cases inspire users to keep their alcohol use down [22]. A very recent content analysis of 800 alcohol apps with a focus on behavior change techniques found that the majority "implicitly or explicitly promoted the use of alcohol". Of 61 apps coded for behavior change techniques, the most frequent techniques used (over 40%) were self-monitoring, information on negative consequences of alcohol use and positive consequences of abstinence, and personalized feedback. The analysis cites no research specifically evaluating the apps reviewed [23].

Indeed, systematic research on mobile phone apps for university students has so far been very sparse [24]. One study targeted smoking and HED based on BASICS (Brief Alcohol Screening and Intervention for College Students) and found no reduction over a one-month period in HED, although students who completed more intervention modules were less likely to drink during the initial 14-day assessment period in the study [25]. In a second study, our own research group conducted a 3-armed randomized controlled trial among university students with problematic drinking, comparing the effects of 2 different apps, one offering personalized feedback on estimated blood alcohol concentration (eBAC) at live drinking occasions and the other offering planning, live monitoring of eBAC with personalized feedback, and follow-up of specific drinking occasions based on eBAC (see Figure 1 for an overview of app components). We found no changes in drinking parameters over a 7-week period apart from an *increase* in number of drinking occasions among men for one of the apps tested. Also, dropping out from the study was associated with drinking at levels higher than the recommended maximum number of standard drinks per week (9 for women and 14 for men); students who drank at higher than recommended levels comprised a sub-group of almost one-third of students assessed as having at least hazardous drinking habits according to the Alcohol Use Disorders Identification Test (AUDIT) [26].

In recent years, alcohol apps for mobile phones have seen an exponential rise in growth. Their potential is high in view of their extensive reach in the population, but the evidence for their effectiveness in reducing problematic alcohol use is lacking [27]. Questions that remain to be answered are whether it is possible to reduce hazardous or even harmful drinking with apps, as well what components an app actually needs to include in order to be effective.

Figure 1. Behavior change components included in the two apps used in Study 1 and in the app used in Study 2.

Behavior Change Components	Promillekoll	PartyPlanner	TeleCoach
Providing information on Protective Behavioral Strategies (PBS)	X	X	X
Planning future parties		X	
Realtime feedback on blood alcohol count (BAC)	X	X	
Comparative follow-up ("How did it go?")		X	
Registering intake over time		(X)	X
Skill training for saying no to alcohol and feeling better without alcohol			X

Study Aim

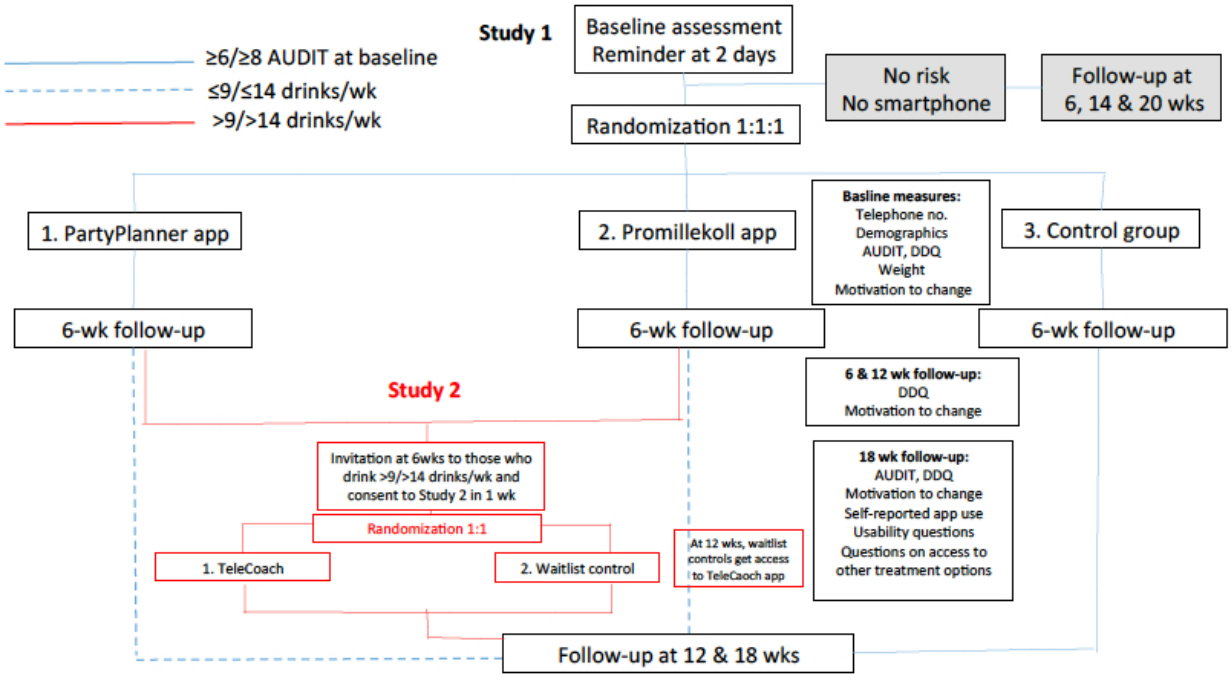
This research has two principal aims. First, we aim to evaluate mobile phone apps in improved, revised versions compared to our previous study [26] for university students with problematic levels of alcohol use based on AUDIT scores for at least hazardous drinking. We also aim to extend follow-up beyond 6 weeks, to 12 and 18 weeks. We hypothesize that the improved app versions may show positive effects with reduced alcohol use over time in comparison to an untreated control group, in contrast to no effects in the 6-week follow-up in our previous study [26]. Secondly, we aim to target the needs of a sub-group of university students with particularly high alcohol consumption according to national public health thresholds for unhealthy drinking, by evaluating a new mobile phone app with skills enhancement functions. We hypothesize that dropout over time will be reduced for students in this sub-group, who are offered an app addressing their specific, more extensive need of help to control drinking. Also, we hypothesize that students offered this app will reduce their drinking to a larger extent than students in a wait-list control group as well as those in an untreated control group (separately culled from the wider data set for Study 1). These aims will be addressed through two consecutive randomized trials (Trial Registration at ClinicalTrials.gov, identifier NCT02064998).

The specific research questions are as follows:

Study 1: Among university students with at least hazardous alcohol use according to the AUDIT, does access and self-reported use of two different mobile phone apps (see Figure 2) lead to reduced alcohol consumption in comparison to an untreated, assessment only control group, at 6, 12 and 18 weeks after registration for the study?

Study 2: Among university students who drink over the recommended 9 and 14 standard drinks per week for women and men, respectively, does access and self-reported use of the TeleCoach app (see Figure 2) lead to lower proportions of individuals with consumption levels under the recommended number of drinks per week, in comparison to a wait-list control group and an untreated control group, 6 and 12 weeks after registration to Study 2 (12 and 18 weeks after registration for Study 1)?

Figure 2. Flowchart showing study design for Studies 1 and 2, including follow-up measures.



Design

We plan to conduct two consecutive randomized controlled studies.

As in our previous trial [26], we plan to cooperate with the student unions at Stockholm University, KTH Royal Institute of Technology and Södertörn University. Students are contacted via direct email or via advertisements on student union websites or Facebook pages. The direct emails provide information to prospective study participants on a randomized study of mobile phone mobile apps concerning alcohol habits. Prospective participants clicking on the invitation link will be asked to fill in information on age, gender, term of study, possible previous use of apps, and questions on alcohol use according to the Daily Drinking Questionnaire (DDQ) [28] and the 10-item AUDIT [29].

Any interested person completing the online registration process will be included in the study, regardless of level of alcohol use. Individuals without any hazardous drinking will thus be recruited for this research including all follow-ups, but they will not be randomized to any intervention. We will conduct a later secondary analysis of all data including students without hazardous drinking, to explore and discuss the regression to the mean phenomenon in this group [30].

Problematic alcohol use will be defined in two ways. All potential participants will have filled in the AUDIT and the DDQ. Those with AUDIT scores of ≥6 for women and ≥8 points for men will be defined as drinking alcohol at levels that are at least hazardous to their health. Those with weekly drinking levels according to the DDQ, of over 9 drinks per week for women and 14 drinks per week for men, will be defined as having elevated alcohol consumption levels. The AUDIT-based definition is the basis for recruitment to Study 1, whereas the DDQ-based definition is the basis for recruitment to Study 2.

Procedure

A flowchart showing the procedure and interconnection between Studies 1 and 2 is shown in Figure 2.

First Study

In Study 1, we basically repeat the design of our original study from spring 2013, in which students with problematic alcohol use were randomly allocated to one of three groups: the Swedish Alcohol Monopoly's Promillekoll app, the research group's PartyPlanner app or a control group. The planned study differs from the original study in two aspects. First, app content will have been updated for both apps (see below under Interventions). Secondly, the follow-up time is extended from only one follow-up at 6 weeks, to three follow-ups at 6, 12 and 18 weeks.

Participants owning a mobile phone and showing at least hazardous use of alcohol, defined as scores of ≥ 6 for women and ≥ 8 for men on the AUDIT, are randomized to a group with access to one of the two apps, or to an untreated, assessment-only control group. Participants randomized to an intervention are sent an email with a link to the respective app with instructions on how to install the app, and a brief recommendation to use the app on drinking occasions. As noted above, participants without risky alcohol use and/or without a mobile phone are excluded from randomization. All participants are informed that they will be contacted after 6, 12 and 18 weeks via email for follow-up.

Second Study

Participation in Study 2 will be offered to students in the Study 1 intervention groups who, at 6-week follow-up, show elevated alcohol consumption in excess of 9 and 14 standard drinks a week according to the DDQ (for women or men, respectively). This level of alcohol consumption is connected to an elevated

risk of harmful consequences, and is of particular concern since 1 out of 4 deaths among individuals 15-29 years old in the EU are alcohol-related [31]. These students are given feedback on their elevated consumption level with higher risk of harmful consequences. They continue to have access to the app they were randomized to in Study 1, but are randomized to one of two groups: access to an in-depth self-help TeleCoach app that the research group has developed, or to a wait-list control group who will be given access to the app at the 12-week follow-up.

All study participants who complete the final follow-up will participate in a lottery of 3 iPad devices, through a partnership with Save the Children.

Interventions

All three app interventions used in Studies 1 and/or 2 are described below. Figure 1 shows a simplified overview of basic behavior change components included in each app.

Promillekoll App (Translation: "Check your BAC")

This mobile phone app is available in iPhone and Android versions, and allows the user to register alcohol consumption in real time, giving immediate feedback on the eBAC. The app alerts the user if s/he surpasses an alcohol concentration of 0.06 percent BAC, a level where negative consequences can begin to occur, and only displays values up to 0.08 percent, in an effort to emphasize the message to users that higher eBAC levels are harmful. The app also provides separate information texts on alcohol and BAC. The app was publicly launched by the Swedish Alcohol Monopoly (Systembolaget) in the autumn of 2012 and improved a year later on the basis of the Monopoly's own surveys and 343 student comments from our study in the spring of 2013 (unpublished data). The Promillekoll app is theoretically based on the assumption that information about one's own real-time eBAC levels can contribute to protective cognitive and behavioral strategies. A further mechanism, congruent with the Theory of Planned Behavior (TPB) [32], is that providing information and feedback on risky levels of eBAC modifies the intention to consume alcohol. Promillekoll also offers selected specific protective behavioral strategies (PBS) [33] to maintain alcohol consumption at or below the 0.06 percent BAC level. No user data are collected.

PartyPlanner App

This Web-based app guides the user to plan drinking occasions so the consumption level stays below risky levels of eBAC, at about 0.06 percent. The app also allows the user to record real-time consumption and provides immediate feedback on how to adjust their consumption to maintain a healthy eBAC level to 0.06 percent or less. If the user has created a plan for a specific drinking occasion and then followed it up, the app offers a graphic visual comparison of the plan, with the logged real-time event after the actual drinking occasion. These app components are in line with the goal setting and personalized feedback components of successful BMIs [9].

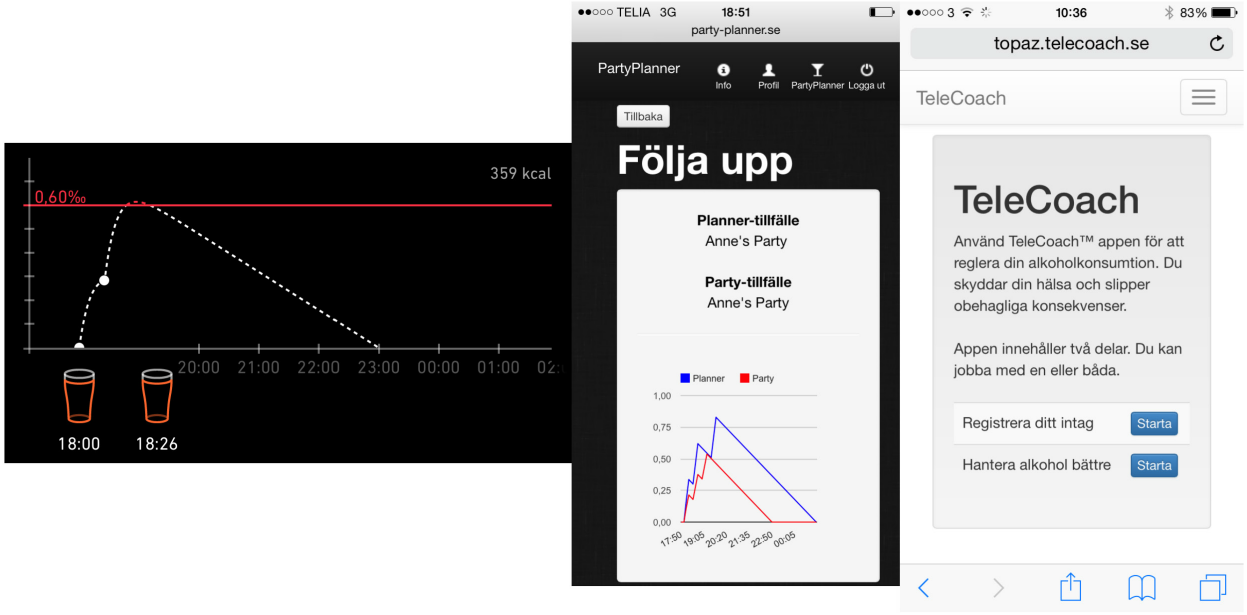
The app was built by the research team in the fall of 2012 and revised in the fall of 2014 for the current study. In the revised version, users receive feedback about the health benefits of lower alcohol use and potential social losses connected to over-consumption, since laboratory studies have shown that this kind of information affects men's drinking intentions in a downward direction [34]; we focused on adapting information primarily to men because we saw a significant negative effect for men using Promillekoll in our first study, and unpublished secondary analyses suggested that men might have more difficulty using an app constructively in a social context where alcohol is consumed, than women. An example of the type of feedback given is shown in Figure 3. PartyPlanner also includes several PBS [33] to maintain alcohol consumption at a minimally harmful level.

Note: The level for non-risky eBAC is below 0.06 percent for both Promillekoll and PartyPlanner, in correlation with guidelines used in the BASICS program [35]; this level is somewhat more conservative than the 0.08 percent level correlated with the concept of binge drinking [36].

TeleCoach App

This app is based on an Interactive Voice Response (IVR) automated telephony intervention developed by the research group in 2009-2012 [37]. Aside from offering skill acquisition for PBS [33], it provides skill training exercises for individuals who want to reduce or end their alcohol consumption but have experienced trouble in achieving these behavioral changes. The app includes a behavioral chart, and modules on "saying no to alcohol" and "feeling better without alcohol." The "saying no" module includes instruction on firm body language and voice, a guide to the five principles of saying no [38] and the Alcohol Abstinence Self-Efficacy Scale [39]; the "feeling better" module includes relaxation exercises, lists of positive thoughts [40] and urge surfing [38]. In this study, we will test the app version of the intervention for the first time, targeting university students who consume alcohol over the national recommendations of no more than 9 and 14 standard drinks/week for woman and men, respectively.

Figure 3. Screen dumps from each of the three apps described in the article. From left to right: Promillekoll, PartyPlanner, TeleCoach.



Assessment

The baseline, 6, 12 and 18 week follow-ups, include the Daily Drinking Questionnaire (DDQ) [28], and all 10 questions in the AUDIT [41]. The DDQ was developed in the US and was adapted for use by students by Collins et al (1985). The instrument was translated and adapted for use in Sweden by the Department of Clinical Alcohol Research at Lund University, and has been used in previous studies of Swedish students at the university [42]. The AUDIT is an effective instrument for identifying problematic alcohol consumption. The instrument was developed by the World Health Organization (WHO) and measures consumption as well as signs of harmful use and dependency related to alcohol use. The Swedish version was translated and disseminated by researchers at Karolinska Institutet [43]. The assessment package also includes information about participants' mobile phone number, gender and age, and possession of a mobile phone. All follow-ups also include questions about users experience interacting with the apps. The 18-week follow-up also includes questions regarding access, during the 18 weeks since registration, to other types of treatment such as additional sources of information, medication, speaking to counselors or using other apps than the one they are randomized to; more information on these questions is available in studies on minimal Web-based intervention conducted by our group [44,45]. Invitations to participate in follow-ups are sent via e-mail, where participants are asked to log on to a secure survey to complete assessments. Two email reminders are sent out during the one-week time period allowed for response.

Power Calculation

Primary outcomes for both studies are quantity, frequency, number of binge drinking occasions as well as mean and peak eBAC, and proportion of individuals drinking over the recommended levels of 9 and 14 drinks per week, for women and men respectively.

For Study 1, in order to answer the questions on whether the use of a mobile phone app affects alcohol consumption compared with controls, 82 individuals need to be included in each of the three groups (two intervention and one control group, 246 individuals in total) in order to detect a difference in effect size 0.10 at 5% -level with a power of 80%. This is based on an assumed correlation between DDQ, pre- and post-measurement of at least 0.5. It is likely, however, that high power can be achieved since approximately 7,000 students will be invited to participate in the study. We expect at least 1200 students to agree to participate based on the 17% recruitment level achieved in our previous study [26], but we hope more will participate since we will be targeting first- and second-term university students in this research, in contrast to all registered students regardless of study level, in the previous study. About half of the recruited students are expected to have at least hazardous use and be eligible for randomization.

For Study 2, based on the same assumption as above, 82 individuals need to be included in each group (intervention and wait-list control group), 164 individuals in total. Based on our prior study [26] we expect that approximately 30% of the study participants will have an elevated risk consumption at 6-week follow-up, and estimates based on previous experiences of similar design [46] suggest that at least 50% will want to participate in the study, suggesting that adequate power will be available.

Ethics and Time Plan

The project was approved by the regional ethical review board on March 19, 2014 (2014/278-31/2). The study commenced in early autumn 2014. Recruitment began during the last week of September, 2014. Participants who at 6-week follow-up showed harmful use of alcohol over 9 or 14 standard drinks per week for women and men, respectively, were offered participation in the second study. All participants were followed up at 6, 12 and 18 weeks after baseline recruitment to the project. Data collection was completed in the late spring of 2015. The results

will be analyzed during the autumn of 2015, see below for the analysis plan.

Results

Studies 1 and 2 will be analyzed separately. For both studies, descriptive statistics will be used to describe baseline characteristics. Analysis of variance (ANOVA) will be used to identify any baseline differences in age, AUDIT, quantity, frequency, number of binge drinking occasions, as well as mean and peak eBAC between the groups. Pearson's chi-square tests will be used to determine differences between the groups in the gender distribution and the proportion of participants drinking more than the weekly recommendation.

For Study 1, all participants in both studies will be included in the analysis. Latent Markov models [47] will be used to maximize differing results for Study 1 participants not offered participation in Study 2, Study 1 participants who declined participation in Study 2, and Study 2 participants.

For Study 2, a linear mixed model analysis will be used to identify changes over time in alcohol consumption outcomes: quantity, frequency, and number of binge drinking occasions, mean eBAC and peak eBAC. These analyses will be conducted per protocol—that is, including only those participants who report using the app they were assigned to, and controlling for self-reported access to other treatment sources for problematic alcohol use during the study, and having accessed the publicly available Promillekoll app prior to the study. For comparison, intention to treat analyses will be performed with all participants randomized to experimental groups and retaining baseline values for as many participants as possible.

Descriptive statistics, ANOVA and Pearson's chi-square analyses will be performed using IBM SPSS Statistics for MacOS X, Version 22 (IBM Corp). Linear mixed model analyses will be performed using Stata 13 (StataCorp). Values for averages and standard deviations are presented to three decimal places for variables where this is necessary in order to make differences visually discernible.

Discussion

Summary

Harmful alcohol use is a significant problem in university students. Even though effective face-to-face interventions are available, most students prefer the digital interventions developed in recent years [48]. Computer use has also undergone rapid changes, and today most interaction with computers is through mobile phones [49]. Literally thousands of alcohol apps have developed for mobile phones, but the evidence for their effectiveness in reducing problematic alcohol use is lacking [21,23,27]. Our research studies seek to test the effectiveness of alcohol apps for mobile phones in reducing problematic alcohol use in university students, as well what components an efficient app actually needs to include in order to be effective.

The present research project, as well as our prior research studies [26,42], is a collaboration with the student unions at the major universities in Sweden. Our research group has over 20 years experience cooperating with student unions in alcohol research, and our overall experience is that student unions are an effective partner offering both contact information to their members, credibility, and the potential for implementing positive research findings.

The two apps our research team developed for this study are based on several years of experiences in developing and testing face-to-face brief interventions, as well as computerized brief interventions using both desktop computers and interactive voice response (IVR).

Pros and Cons

In the present project, we decided to use Web-based apps for the two applications our research team developed. This decision has both pros and cons, as Web-based apps are cheap, easy to develop and store user data that is immediately available on our own server, lowering the risk of data losses that may occur when using regular apps run on the mobile phone itself. Regarding cons, users may perceive Web-based apps as a bit slow and sometimes absent, as they are dependent on Internet access. These cons are especially important when comparing the two Web-based apps to a regular app developed by the Swedish retailing company concerning for instance user satisfaction.

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Authors' Contributions

Authors AHB, CA and MG conceived the study design. Authors AHB, CA, and MG revised the content of the PartyPlanner and TeleCoach apps, and MF was responsible for all technical development. MG and AHB were responsible for data collection. KS and AHB conceived the analysis plan. AHB wrote the first manuscript draft. The final draft integrated comments from KS and was written by AHB and CA.

Conflicts of Interest

None declared.

Multimedia Appendix 1

Peer review report from funder (in Swedish).

[PDF File (Adobe PDF File), 78KB - resprot_v4i4e139_app1.pdf]

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Abbreviations

AUDIT: Alcohol Use Disorders Identification Test
BAC: blood alcohol concentration
BASICS: Brief Alcohol Screening and Intervention for College Students
BMI: brief motivational intervention
DDQ: Daily Drinking Questionnaire
eBAC: estimated blood alcohol concentration
HED: heavy episodic drinking
IVR: interactive voice response
PBS: protective behavioral strategies
SBI: screening with brief intervention
WHO: World Health Organization

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Original Paper

The Effect of Patient-Specific Cerebral Oxygenation Monitoring on Postoperative Cognitive Function: A Multicenter Randomized Controlled Trial

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Abstract

Background: Indices of global tissue oxygen delivery and utilization such as mixed venous oxygen saturation, serum lactate concentration, and arterial hematocrit are commonly used to determine the adequacy of tissue oxygenation during cardiopulmonary bypass (CPB). However, these global measures may not accurately reflect regional tissue oxygenation and ischemic organ injury remains a common and serious complication of CPB. Near-infrared spectroscopy (NIRS) is a noninvasive technology that measures regional tissue oxygenation. NIRS may be used alongside global measures to optimize regional perfusion and reduce organ injury. It may also be used as an indicator of the need for red blood cell transfusion in the presence of anemia and tissue hypoxia. However, the clinical benefits of using NIRS remain unclear and there is a lack of high-quality evidence demonstrating its efficacy and cost effectiveness.

Objective: The aim of the patient-specific cerebral oxygenation monitoring as part of an algorithm to reduce transfusion during heart valve surgery (PASPORT) trial is to determine whether the addition of NIRS to CPB management algorithms can prevent cognitive decline, postoperative organ injury, unnecessary transfusion, and reduce health care costs.

Methods: Adults aged 16 years or older undergoing valve or combined coronary artery bypass graft and valve surgery at one of three UK cardiac centers (Bristol, Hull, or Leicester) are randomly allocated in a 1:1 ratio to either a standard algorithm for optimizing tissue oxygenation during CPB that includes a fixed transfusion threshold, or a patient-specific algorithm that incorporates cerebral NIRS monitoring and a restrictive red blood cell transfusion threshold. Allocation concealment, Internet-based randomization stratified by operation type and recruiting center, and blinding of patients, ICU and ward care staff, and outcome assessors reduce the risk of bias. The primary outcomes are cognitive function 3 months after surgery and infectious complications during the first 3 months after surgery. Secondary outcomes include measures of inflammation, organ injury, and volumes of blood transfused. The cost effectiveness of the NIRS-based algorithm is described in terms of a cost-effectiveness acceptability curve. The trial tests the superiority of the patient-specific algorithm versus standard care. A sample size of 200 patients was chosen to detect a small to moderate target difference with 80% power and 5% significance (two tailed).

Results: Over 4 years, 208 patients have been successfully randomized and have been followed up for a 3-month period. Results are to be reported in 2015.

Conclusions: This study provides high-quality evidence, both valid and widely applicable, to determine whether the use of NIRS monitoring as part of a patient-specific management algorithm improves clinical outcomes and is cost effective.

Trial Registration: International Standard Randomized Controlled Trial Number (ISRCTN): 23557269; http://www.isrctn.com/ISRCTN23557269 (Archived by Webcite at http://www.webcitation.org/6buyrbj64)

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KEYWORDS

cardiac surgery; cardiopulmonary bypass; transfusion; cognition; valve; coronary artery; asepsis; infection; randomized clinical trial; cerebral oxygenation

Introduction

The Clinical Problem

The development of cardiopulmonary bypass (CPB) represents one of the greatest achievements of modern medicine. By maintaining adequate perfusion through an extracorporeal pump and oxygenator, CPB enables complicated surgical procedures to be performed on the heart. Since its development in the 1950s, refinement of CPB technology has made cardiac surgery safer and widened its availability to increasingly elderly and sicker patients. Perfusion technology remains imperfect, however, and CPB is still associated with significant morbidity [1]. The pathophysiology of CPB-associated morbidity is multifactorial but includes regional hypoperfusion and tissue hypoxia, often within vascular beds, which are already abnormal due to advanced age or comorbidities such as diabetes. Consequently, up to 25% of patients may experience neurocognitive decline [2] and up to 25% experience acute kidney injury [3].

Oxygen Delivery During CPB

Adequate tissue oxygen delivery during CPB is achieved through the optimization of several parameters including CPB pump flows, perfusion pressure, hematocrit, and the oxygen saturation of arterial blood. In contemporary clinical practice in adult cardiac surgery, the adequacy of perfusion is determined by the following: (1) the use of global measures of oxygen utilization such as the mixed venous oxygen saturation (SvO_2), or (2) evidence of tissue hypoxia as implied by elevated serum lactate, a marker of anaerobic cell metabolism, or other indicators of metabolic acidosis. However, in some patients, particularly those with pre-existing end organ dysfunction, global measures may not detect regional hypoxia [4-7].

Direct measures of regional tissue oxygen levels such as gastric tonometry, laser Doppler flowmetry of the intestinal mucosa, or cerebral venous oxygen saturation using jugular bulb catheters can be used to measure and optimize tissue oxygenation. However, these modalities are invasive which limits their use. More recently, near-infrared spectroscopy (NIRS) has emerged as a noninvasive and accurate technique to monitor regional tissue oxygenation [8-12]. An added benefit is that NIRS can accurately measure tissue oxygenation in the brain, one of the most important end organs. NIRS sensors, when applied to the forehead, can determine the relative saturation/desaturation of blood within cerebral arterioles and venules. NIRS cerebral oximetry has been clinically validated as a measure of cerebral venous oxygen saturation [11,13,14], and is now approved as a noninvasive measure of regional cerebral oxygenation by the Medicines and Healthcare Products Regulatory Agency (MHRA). Several observational and a small randomized trial in adult cardiac surgery [8-10,12] have shown that optimization of cerebral oxygenation using NIRS can be associated not only with reduced neurological morbidity but also with a reduction in renal complications and other major adverse clinical events. The wider benefit from optimization of cerebral oxygenation observed in these studies may have arisen because the brain is more susceptible to hypoxia than other organs [4,5,7].

Whereas cerebral oximetry is widely used in pediatric cardiac surgery, the relatively low level of clinical evidence supporting its use in adult cardiac surgery has led to wide variation in its adoption, particularly as it carries additional disposable costs of up to £200 per patient. There is thus a need for high-quality evidence to address the benefits and costs of NIRS cerebral oximetry in adult cardiac surgery patients.

Indications for Red Blood Cell Transfusion During Cardiac Surgery

Cerebral oximetry may also be used during CPB to develop goal-directed, patient-specific indicators of the need for red blood cell (RBC) transfusion if used as part of a wider algorithm designed to optimize cerebral oxygenation. The primary goal of RBC transfusion is to optimize tissue oxygenation. Currently, most RBC transfusions are given solely as a response to a hematocrit that has fallen below an arbitrary threshold and not as a response to incipient tissue hypoxia [15-17]. However, the hematocrit is a poor indicator of regional tissue hypoxia, and therefore is inadequate as an indicator of the need for transfusion.

The hematocrit below which oxygen delivery to tissues is reduced such that anaerobic metabolism occurs is known as the “critical hematocrit (Hct).” In healthy human adults very low hematocrits of less than 15 can cause organ hypoxia [5,7], and transfusion in such cases is known to be beneficial [18,19]. However, the Hct for patients during cardiac surgery is unclear; it is thought to be higher than 15, as most patients are elderly and have comorbid conditions, but it is also likely that the Hct varies considerably both between cardiac patients and individual patients over the course of the perioperative period [20].

Hct is increased by conditions which impair autoregulation such as diabetes or increased age [21,22], and during CPB. Hct is also affected by multiple factors that affect the balance of oxygen supply and demand such as hypothermia [23], rewarming [24], pump flow [25,26], or perfusion pressure [25,27]. Uncertainty about when an individual may benefit from

a transfusion is reflected by the wide range in reported hematocrit transfusion thresholds used during CPB (from 17 to 25) [28-30], as well as the wide variation in transfusion rates in cardiac surgical patients across units in the UK (35-75%) [31]. Because the Hct for any particular patient at any given time is unknown, the ability to directly measure tissue oxygenation and incipient tissue hypoxia may have distinct advantages over the use of generic and prespecified hematocrit transfusion thresholds.

Risks Associated With Red Blood Cell Transfusion

Arbitrary, generic transfusion thresholds can result in patients being undertransfused and overtransfused. In cardiac surgery, unnecessary RBC transfusion has been shown in observational studies to be associated with increased rates of stroke, renal dysfunction, infection, prolonged ventilation times, and death [32-34], although this was not confirmed in a recently reported TITRe2 randomized trial [35]. Irrespective of the effects of transfusion on postoperative morbidity, preventing unnecessary blood transfusion would reduce health care costs. Our goal is to develop transfusion algorithms that are objective, patient specific, and directed at optimizing tissue oxygenation and the adequacy of oxygen delivery. We propose that RBC transfusion should be more appropriately given in response to an objective measure of regional tissue oxygenation, as one of a group of interventions, rather than as a response to a generic, prespecified hematocrit threshold in isolation.

Cerebral Oximetry

Existing protocols to optimize tissue oxygenation during CPB deploy manipulations of CPB (according to levels of blood markers such as lactate) and the transfusion of RBC (according to the hematocrit). These methods are used independently of each other and in response to generic thresholds, despite the fact that both have the underlying aim of improving tissue oxygenation.

In this trial we compare alternative algorithms for optimizing tissue oxygenation during CPB (ie, a currently used generic algorithm versus a patient-specific algorithm). The patient-specific algorithm differs from the generic algorithm in two key ways. First, it considers the option to give an RBC transfusion as one element of several linked interventions to optimize regional tissue oxygenation, rather than in response to a predefined hematocrit threshold in isolation. Second, it is "goal-directed" such that the algorithm is specifically targeted to maintain cerebral oxygen delivery during CPB (monitored by NIRS).

The patient-specific algorithm aims to optimize the cerebral oxygen supply and demand balance during CPB by (1) increasing oxygen supply using hyperoxygenation, increased pump flow, perfusion pressure, or hypercapnic cerebral vasodilation; (2) increasing oxygen offloading by the use of nitrates; or (3) reducing oxygen demand by deepening anesthesia [10]. Cerebral oxygen saturations approaching a low threshold in the presence of anemia (a hematocrit of 18-23) and despite optimization of other parameters suggest that the cerebral Hct is about to be reached and transfusion is indicated. Therefore, this algorithm is patient and time specific, and goal-directed to optimize a validated objective measure of tissue oxygenation. This could potentially reduce health care costs associated with unnecessary allogeneic RBC transfusions and complications associated with tissue hypoxia during CPB.

Aims and Objectives

We aim to compare generic and patient-specific algorithms for optimizing tissue oxygenation during CPB in adult cardiac surgery patients in a randomized controlled trial (RCT). Compared to the generic algorithm (including a hematocrit transfusion threshold of 23), we hypothesize that the patient-specific, goal-directed algorithm (based on optimizing regional cerebral oxygen saturation), combined with a prespecified "restrictive" hematocrit transfusion threshold of 18, will result in fewer RBC transfusions and will reduce complications arising from low oxygen levels during CPB. The specific objectives of this multicentre RCT are to (1) compare the effects of the patient-specific, goal-directed algorithm versus the generic algorithm in terms of cognitive function; (2) compare the effects of the patient-specific, goal-directed algorithm versus the generic algorithm with respect to a range of secondary outcomes; and (3) estimate the cost effectiveness of the patient-specific, goal-directed algorithm versus the generic algorithm and describe this in terms of a cost-effectiveness acceptability curve.

Methods

This is a multicenter, parallel group RCT of two different algorithms for optimizing tissue oxygenation in adult cardiac surgical patients undergoing nonemergency valve surgery, with or without coronary artery bypass graft (CABG), and using CPB at mild hypothermia (32-35°C). The trial is restricted to these patient groups because they have a higher chance of needing transfusion and needing more RBC units transfused than patients undergoing isolated CABG (Trial Registration Number [ISRCTN]: 23557269).

Trial Population and Recruitment Procedure

The target population is adult cardiac surgical patients undergoing nonemergency valve or combined CABG and valve surgery using CPB. Patients may enter the trial if they are adults (≥16 years) of either sex undergoing valve or combined CABG and valve surgery at the Bristol Royal Infirmary in Bristol, Glenfield Hospital in Leicester, or Castle Hill Hospital in Hull, give informed consent, and are suitable for allocation to either transfusion protocol. Patients are excluded from the trial if any of the exclusion criteria apply (Textbox 1).

Textbox 1. Exclusion criteria for the trial.

- Patients undergoing emergency cardiac surgery.
- Patients who are prevented from having blood and blood products according to a system of beliefs.
- Patients who may have higher perioperative hemoglobin requirements or critical limb ischemia.
- Patients with congenital or acquired RBC, platelet, or clotting factor disorders.
- Patients with a neurological disorder.
- Patients with a diagnosed psychiatric disorder, drug, or alcohol addiction.
- Patients with an already identified cognitive impairment as defined by psychometric assessment or a preoperative Mini Mental State Examination score of <24 [36].
- Patients who have previously sustained a stroke, intracerebral hemorrhage, or acquired brain injury.
- Patients with a pre-existing inflammatory state.
- Patients with end-stage renal failure or patients who have undergone renal transplantation.
- Patients unable to complete the cognitive assessments required for the trial (eg, due to language difficulties, visual, or hearing impairment).
- Patients who are unable to give full informed consent for the study.
- Patients already participating in another clinical (interventional) study.

Potential trial participants are identified from surgical theatre lists and sent study information by mail or fax. They are approached on admission for surgery and after written consent has been obtained and the patient has scored 24 or greater on the Mini Mental State Examination (MMSE), he/she is randomized by a member of the research team prior to surgery. Confirmation of a participant's identity and eligibility has to be entered into a database before the randomized allocation is generated. Some consented patients are found to be ineligible because they have a MMSE score of less than 24. Details of all patients approached for the trial and reason(s) for nonparticipation (eg, reason for being ineligible, patient or clinician preference, or patient refusal) are documented.

Randomization

Participants are randomized in blocks of varying size in a 1:1 ratio, stratified by center and planned surgery (valve only or combined CABG and valve). Allocations are generated by computer in advance of starting recruitment and are concealed using an Internet-based system. To maintain blinding, a member of the staff who is not involved in data collection, neurocognitive assessment, or providing health care randomizes participants shortly before surgery. The allocation is given to the responsible clinical perfusionist in a sealed envelope by the trial coordinator. If surgery is unexpectedly rescheduled, participants retain their study numbers and allocation. Eligible patients who consent to participate and who have an MMSE score of 24 or greater are randomly allocated to one of two different algorithms for optimizing tissue oxygenation during CPB (Textbox 2).

Textbox 2. Algorithms for optimizing tissue oxygenation.

- "Generic algorithm" (including a standard transfusion threshold): Based on global measures of oxygen utilization and includes a predefined intraoperative hematocrit transfusion threshold of 23 (current standard practice, the existing protocol).
- "Patient-specific algorithm" (including a restrictive transfusion threshold): Patient-specific, goal-directed algorithm based on the monitoring and optimization of regional cerebral oxygen saturation, combined with a predefined restrictive intraoperative hematocrit transfusion threshold of 18.

For patients allocated to the patient-specific algorithm, optimization of cerebral oxygenation uses a protocol based on that suggested by Murkin and colleagues [8]. Target regional oxygen saturation values are specified as 70% or more of preinduction values and an absolute value of 50% or more. However, if the regional oxygen saturation remains at or above these target values, but the hematocrit falls to 18, then RBC transfusion is indicated (this represents a measure to increase patient safety and protect participants from possible risks associated with the experimental intervention). Several measures are taken to maintain these targets (Table 1).

Table 1. Cerebral oxygenation optimization protocol to maintain cerebral oximetry readings within 70% of baseline and greater than 50%.

Measure	Description
Equipment check	Bypass pump, oximeter sensors, head and aortic cannula position
Cerebral perfusion pressure[a] (CPP) <60 mmHg	Raise CPP > 60 mmHg by administering 0.5 mg metaraminol
If no effect and CPP < 80 mmHg	Raise CPP > 80 mmHg by administering 0.5 mg metaraminol
Partial pressure of CO_2 in arterial blood ($PaCO_2$) < 35 mmHg	Raise $PaCO_2$ to 40-45 and reduce gas flows
Inspired oxygen concentration (FiO_2) < 0.6%	Raise FiO_2
If no effect and FiO_2 < 1.0	Raise FiO_2 to 1.0
Decrease cerebral metabolic rate	Increase depth of anesthesia by increasing the propofol infusion rate
Increase pump flow to maximum tolerated by venous reservoir	
Target not met and critical hematocrit is 18-23	Transfuse 1 unit of RBC

[a]CPP=mean arterial pressure (MAP)-central venous pressure (CVP).

Clinical Management Protocols

The following protocols are followed for all patients unless otherwise specified.

Surgery and Anesthesia

Details of anesthesia, operation type, complexity, and postoperative management are recorded. To avoid confounding by variables known to affect regional perfusion (eg, anesthetic depth, pCO_2, crystalloid infusions) anesthetic management will adhere strictly to existing protocols [37], except where required according to the individual, goal-directed protocol for patients randomized to the patient-specific algorithm (Table 1).

Cardiopulmonary Bypass

All patients are managed according to a standard CPB protocol for valve surgery using mild hypothermic CPB and intermittent antegrade/retrograde cold blood cardioplegic arrest as previously described [37]. All distal anastomoses are conducted during a single period of aortic cross clamp. CPB pump flows, hematocrit, and arterial and mixed venous oxygen saturation are recorded every 20 minutes according to standard practice. In addition, cerebral oximetry is continuously measured in all patients for the duration of the operative procedure (see the following section).

Near-Infrared Spectroscopy Monitoring of Cerebral Oxygenation

Regional oxygen saturation of blood in the cerebral cortex is measured using the INVOS 5100 regional oximeter (Somanetics, Troy, MI, USA) [10]. NIRS readings are recorded for patients in both study arms for comparison. Bilateral NIRS sensors are attached to the patient's forehead prior to preoxygenation and anesthetic induction to calculate baseline values. Cerebral oximetry is continuously measured and recorded for the duration of the operative procedure and discontinued once the patient has left the operating theater.

For patients randomized to the patient-specific algorithm (intervention group), the protocol for perioperative optimization of cerebral oxygenation/transfusion (described in Table 1) is performed. The list of interventions used to optimize cerebral oxygenation is logged. For patients randomized to the generic algorithm (control group), NIRS measurements are recorded for comparison but the clinical team is blinded to these readings in theater and optimization of tissue oxygenation is based on global measures of oxygen utilization only (ie, current standard procedure). For patients in both groups, on return to the cardiac intensive care unit (CICU), the clinical team caring for the patient is blinded to the allocation and the NIRS readings from the operative period.

Transfusion Protocols

RBC transfusion protocols for patients randomized to the patient-specific algorithm and generic algorithm are as described above. One unit of RBC should be transfused and the cerebral oxygenation and hematocrit levels are checked before transfusing another unit.

Clinicians are allowed to transfuse, or refuse to transfuse, in contravention of the allocated transfusion protocol but must document the reason(s) why on the study case report form (CRF). The time and volume of the transfusion administered (if applicable) will be recorded for both groups. Non-RBC blood products will be transfused using existing unit protocols [38].

Primary Outcome

The patient-specific algorithm is designed to both prevent regional (cerebral) hypoxia and reduce the likelihood of a patient having an unnecessary RBC transfusion, compared to the generic algorithm. The primary outcome, cognitive function measured at 3 months after surgery, was chosen to measure the hypothesized benefits of preventing regional (cerebral) tissue hypoxia.

Cognitive function is assessed by a qualified examiner blinded to treatment allocation preoperatively, between 4 and 7 days postoperatively, and again at 3 months. The recommended tests [39] are performed in a fixed order (Textbox 3). In addition, to help interpret the cognitive function data, assessments related to the cognitive testing are carried out for all participants (Textbox 4).

Textbox 3. Cognitive domain tests and order.

- Attention (first trial of the Rey Auditory Verbal Learning Test [RAVLT], and sustained and divided attention; Trail-Making Test parts A and B [40,41])
- Verbal memory (RAVLT [40,42])
- Visuospatial (block design from the Wechsler Adult Intelligence Scale-Revised (WAIS-R) test [43])
- Psychomotor speed (digit symbol test from the Wechsler Adult Intelligence Scale-Revised (WAIS-R) test [43])
- Executive function/verbal fluency (Controlled Oral Word Association Test [COWAT] [44])
- Motor coordination (Grooved Pegboard Test, dominant and nondominant hand [40])

Textbox 4. Assessments related to the cognitive testing.

- The Wechsler Test of Adult Reading provides a preoperative measure of intellectual ability [45].
- Documentation of medications known to interfere with neuropsychological functions (including hypnotics, sedatives, neuroleptics, anxiolytics, antidepressants, and β-blockers) preoperatively, 4-7 days postoperatively, and 3 months postoperatively.
- Assessment of patient's current mental health using the General Health Questionnaire (GHQ-30) and Hospital Anxiety and Depression Scale (HAD) [46] preoperatively, 4-7 days postoperatively, and 3 months postoperatively, to take into account the potential interaction between postoperative cognition and mood.

Secondary Outcomes

Several secondary outcomes are characterized from the collected data (Textbox 5). Systemic inflammatory response syndrome (SIRS) is central to the diagnosis of infective complications and is defined as greater than or equal to two of the following conditions: (1) temperature over 38°C or below 36°C, (2) heart rate greater than 90 beats/minute, (3) respiratory rate greater than 20 breaths/minute or $PaCO_2$ level less than 32 mmHg, and (4) white blood cell count greater than 12,000/mm^3 or less than 4000/mm^3. Blood test results and temperature are classified using standard reference ranges.

Textbox 5. Secondary outcomes.

- Units of RBC and other blood components transfused during the operative period and postoperative hospital stay are recorded.
- Cerebral oxygenation during the operative period: NIRS readings are recorded for both groups for comparison. Monitoring starts before preoxygenation and anesthetic induction and continues until the patient leaves the theater.
- Oxygen delivery and utilization during CPB: serial measurements of oxygen delivery and utilization are collected from the clinical perfusion record.
- EuroQol EQ-5D-3L (a generic health related quality of life instrument that measures mobility, self-care, usual-care, pain/discomfort, and anxiety/depression): assessed at baseline and at 6 weeks and 3 months after surgery.
- Length of CICU or high dependency unit (HDU) stay.
- Length of postoperative hospital stay.
- Clinical outcomes defined as infectious complications (sepsis and wound infection), stroke (validated by CT scanning), ST elevation myocardial infarction accompanied by troponin >5 ng/mL, postoperative acute kidney injury (defined as AKIN criteria stage 1, 2, or 3 [47]), and respiratory complications (ie, reintubation, ventilation >48 hours, tracheostomy, or acute respiratory distress syndrome).
- Sepsis is defined when antibiotic treatment is required for suspected infection and the presence of systemic inflammatory response syndrome (SIRS) within 24 hours prior to start of antibiotic treatment. Sepsis occurring postdischarge only contributes if the event results in admission to hospital or death; wound infection is considered in the following scenario: ASEPSIS score >20, sternum, and if applicable, leg and arm. Wounds will be assessed at least once during a participant's hospital stay. A questionnaire will be administered at 3 months to identify wound infections arising after discharge.
- For stroke diagnosis, blinded assessment of brain imaging (CT or MRI), in association with new onset focal or generalized neurological deficit (defined as deficit in motor, sensory, or co-ordination functions) will be performed.
- Cumulative resource use, cost, and cost effectiveness.
- All-cause mortality within 30 days of surgery.
- Biochemical markers of organ injury.

The following are measured from venous blood samples taken preoperatively, on return to CICU, and 6, 24, 48, and 96 hours postoperatively: (1) S100/100B (brain), (2) troponin I or T (heart), (3) creatinine clearance derived from serum creatinine

(kidney), and interleukins (systemic inflammation). The use of these markers has been described previously [48,49]. Urinary creatinine and electrolytes, urinary microalbumin, neutrophil gelatinase-associated lipocalin (NGAL), interleukin 18 (IL18), liver-type fatty acid-binding protein (LFABP), and kidney injury molecule-1 (KIM-1) [50,51] (markers of tubular and glomerular renal injury) are measured from urine collected for over a 3-hour period (1 sample taken preoperatively and 3 samples taken over the first 2 postoperative days).

Sample Size

The cognitive outcomes are continuously scaled so target differences can be specified as "standardized differences" (0.2 is small; 0.5 is moderate; and 0.8 is large) [52]. In order to detect a small to moderate target difference with 80% power and 5% significance (two tailed), 200 patients (100 per group) are recruited. Specifically, this sample size allows the trial to detect standardized differences between groups of 0.33 (0.36 at 2.5% significance) in cognitive function (adjusting for baseline) and 0.46 in cost, while allowing for up to 12% dropout across the trial. Correlations between measures are assumed to be 0.5 between baseline and postintervention measures and 0.7 between repeated postintervention measures.

This sample size also allows a standardized difference of 0.3 to be detected for biochemical markers (adjusting for baseline and using 4 repeated measures). With respect to CICU and hospital discharge, the sample size allows a hazard ratio of at least 1.65 to be detected.

Statistical Analysis

The primary and other continuous outcomes are analyzed by regression modeling, transforming logarithmically if required, and jointly modeling baseline values where available using mixed models for repeated measures. Serial CPB oxygen delivery and utilization are compared between the 2 groups to determine whether this is improved using NIRS. Time to CICU and hospital discharge is analyzed by Cox regression.

The findings are reported as effect sizes with 95% confidence intervals. Frequencies of complications, which are too infrequent for the trial to be able to detect differences between groups, are tabulated descriptively in accordance with guidelines for reporting RCTs [53]. Analyses are based on intention-to-treat and are adjusted for center and surgery (valve or CABG and valve). For the study to conclude that the patient-specific algorithm is superior to the generic algorithm evidence of superior cognitive function for at least one domain at the 2.5% level is required (the significance level chosen to reduce the likelihood of a type 1 error, but not to be as conservative as a Bonferroni correction).

Health economic analysis are undertaken by the Health Economics Research Centre, University of Oxford. Analyses are designed and conducted to estimate the costs and likely cost of the alternative approaches identified in this protocol. These analyses help to determine whether there are likely to be improvements in patient health outcomes (such as morbidity through complications) associated with the proposed new algorithm, and whether better use can be made of scarce National Health Service (NHS) resources.

Resource use data collection are integrated, where possible, into study documentation supplemented by clinical and laboratory observational data. The analysis calculates the average cost and outcome on a per-patient basis and from this the incremental cost-effectiveness ratios for the different blood algorithms are derived, producing an incremental cost per-complication avoided. Probabilistic sensitivity analysis are used to demonstrate the impact of the variation around the key parameters in the analysis of the baseline cost-effectiveness results. This examines the impact of changing resource use in particular to help with generalizing the study results to other UK settings. The results are expressed in terms of a cost-effectiveness acceptability curve. A single analysis is performed at the completion of the study and no interim analyses and subgroup analyses are planned.

Ethical Approval

The South West Research Ethics Committee approved the trial protocol on June 15, 2009.

Adverse Events

Serious and other adverse events are recorded and reported in accordance with the International Conference for Harmonisation of Good Clinical Practice (ICH GCP) guidelines and the Sponsor's (University Hospitals Bristol NHS Foundation Trust) Research Related Adverse Event Reporting Policy. Castle Hill and Glenfield Hospital will notify the coordinating center (Clinical Trials and Evaluation Unit, Bristol) of all serious adverse events. Data on adverse events are collected from the time of surgery for the duration of the participant's postoperative hospital stay and for the 3-month follow-up period.

Measures to Reduce the Risk of Bias

Concealed randomization prevents selection bias. To assess protocol compliance, interventions made as part of the patient-specific algorithm are logged. NIRS cerebral saturation monitoring is also conducted on patients allocated to both the generic and patient-specific groups to document whether the intervention resulted in significant separation of mean NIRS values. The NIRS values are carefully concealed from the theater team in the generic group. Surgical, anesthetic, and perfusion staff in the theater cannot be blinded to allocation but every effort is being made to ensure that patients, CICU, and other ward care staff and outcome assessors are blinded to minimize performance and detection bias. To further minimize detection bias, outcome measures are defined on the basis of objective criteria. Data are collected for all patients during surgery to characterize compliance with the randomly allocated transfusion protocol. Analysis is on the basis of intention-to-treat.

Dissemination

The findings are disseminated by usual academic channels (ie, presentations at international meetings), as well as by peer-reviewed publications and through patient organizations and, where available, newsletters to patients. As the study compares surgical techniques there will be no commercially exploitable findings from this study.

Results

Patients have been successfully recruited over a 5-year period. Follow-up on all study patients was completed in April 2014. Results are to be published early 2016.

Discussion

Key Changes to the Protocol

There have been changes to the protocol since it was first approved in 2009. The study was designed with co-primary outcomes of cognitive function and infectious complications, to measure both the hypothesized benefits of preventing regional (cerebral) tissue hypoxia and a reduction in unnecessary red blood cell transfusion. Evidence available at that time suggested that RBC transfusion was associated with increased rates of infection. However, the TiTRe2 trial [35] found consistent rates of infection with both liberal and restrictive postoperative transfusion thresholds (92% versus 53% transfused, 25% infection rate in both groups with less than 20% due to sepsis). As a result, infective complications was removed as a co-primary outcome and added as a secondary clinical outcome. Similarly, a cumulative infection score, which we had intended to develop using data from the TITRe2 trial by supplementing data on wound infections with data describing the severity of sepsis was dropped. Instead the occurrence of an infectious complication is reported. There have also been changes to the study biomarkers. Urine levels of LFABP and KIM-1 replaced cystatin C and complement activation in blood samples as more sensitive and specific biomarkers for early detection of glomerular and tubular renal injury. Glutathione-*S*-transferases alpha and pi, novel markers of tubular and glomerular renal injury, were also added but have since been removed after preliminary laboratory test results indicated technical problems with the ELISA kits. The manufacturer was informed of these findings. The blood and urine samples collected were stored for analysis at the end of the trial. The indicated changes have not impacted the integrity of the trial.

The study was initially designed as a single-center study in Bristol. To increase recruitment rates, after 2.6 years, the study was extended to 2 further centers. At the request of the funder, the sample size was reviewed after 100 participants were recruited. The correlation between baseline and postintervention cognitive function was lower than anticipated when the study was planned (0.5 rather 0.7) and there was a higher than anticipated dropout rate early on in the trial (see the following section). To account for these differences, the target sample size was increased from 150 to 200 participants. The target effect size was unchanged.

Protocol Compliance

Perfusion Protocol

Perfusionists involved in the conduct of the trial are experienced in using NIRS equipment due to its routine practice in pediatric cardiac surgery. However, this trial involves the implementation of a specific and more complex NIRS protocol in addition to the routine clinical duties. Therefore, compliance of this protocol is carefully monitored. For each participant, the trial coordinator sets up the NIRS equipment in an anesthetic room and highlights the procedures involved in the generic and patient-specific algorithms prior to handing over the treatment allocation in a sealed envelope. Training sessions and meetings are arranged throughout the conduct of the trial. In order to develop an effective relationship with the perfusionist team the same 3 coordinators are involved with the set up of the NIRS equipment.

Blood and Urine Sample Collection

This trial involves the collection of urine samples at 4 time points and blood samples at 6 time points. These samples are predominately taken by ward staff and analyzed by scientists who have no other involvement in the trial. To make sure all samples are taken in a timely fashion we hold regular teaching sessions with ward staff and have created a sample checklist for them to complete. Research nurses and trial coordinators visit the wards throughout the day to check that all samples are up to date. To reduce the likelihood of sample misplacement, an electronic sample log linked to the freezer where the study samples are stored was developed to indicate the exact location of each study sample within the freezer.

Challenges to Recruitment and Retention of Participants

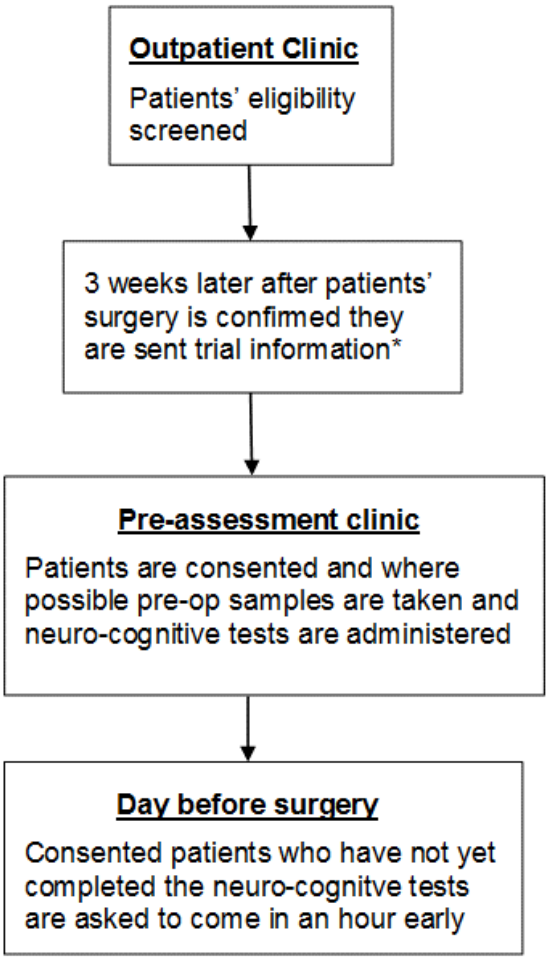
Since the protocol was written a few measures have been introduced to help with the recruitment and retention of trial participants.

Recruitment

When recruitment began, potential patients were identified from operating lists and then sent information by mail or fax. Approach and consent took place once patients arrived in the hospital for their operation. Short periods between patients receiving trial information and operation dates resulted in a number of patients refusing consent. During the course of the study, the Bristol Heart Institute introduced a day of surgery admission (DOSA) policy to maximize bed space. DOSA patients attend the hospital for a short visit the day before surgery to give consent for their procedure and undertake routine clinical tests. They then return for their surgery the following day. Around 50% of patients are now DOSA, thus limiting the amount of time to enroll patients on to the trial, conduct neurocognitive assessments, and take trial samples. To help with recruitment, ethical approval was obtained to approach and consent patients at the preassessment clinic. The pathway that most of our elective patients now follow is shown in Figure 1.

Since approaching patients earlier in their care pathway, only 3.3% (4/120) of eligible patients have refused consent due to insufficient time to consider the study compared to 11% (8/70) who previously refused on these grounds.

Figure 1. Patient recruitment pathway since February 2011.



Retention

The trial requires patients to undergo neurocognitive tests at 3 time points: preoperatively, 5 days after surgery, and 3 months after surgery. These tests can be taxing and each assessment takes up to 90 minutes. As such, patients can become anxious about their performance and this may result in them refusing to complete all of the assessments. For consistency, all staff at the participating sites need to administer tests in the same way. All research team members are trained by the same chartered neuropsychologist and clinical trial co-ordinator. A core part of the training centers on an awareness of patient sensitivity. Staff are assessed and monitored throughout recruitment period.

After discharge, patients may decide they do not wish to come back to the hospital for the 3-month follow-up assessments because they may find it too stressful or may not want to travel. Therefore, where possible, we try to schedule the follow-ups with routine postoperative hospital outpatient appointments. In May 2011, ethical approval was obtained to conduct 3-month follow-up visits at patients' homes. Since home visits have taken place, only 1.7% (2/120) of patients have refused to take part in the follow-up compared with 7% (5/70) previously. Putting these strategies in to place has helped us increase recruitment in to the trial, reduce patients lost to follow-up, and collect high-quality outcome data.

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Authors' Contributions

All authors have read and approved the final manuscript. GM, the Chief Investigator, conceived the trial and participated in the protocol design. BR and CR designed the protocol, edited the manuscript for key intellectual content, and will coordinate and perform the statistical analysis. RD and EN participated in the protocol design and specified the perfusion procedures. SS is the trial Principal Investigator in Bristol. LE drafted the manuscript, coordinated the trial, and administered the neurocognitive assessments. LD has assisted in the trial coordination. GC is the trial statistician who has been involved in the development of the statistical analysis plan and will carry out the final analyses.

Conflicts of Interest

None declared.

Multimedia Appendix 1

NHS Peer review 1.

[PDF File (Adobe PDF File), 137KB - resprot_v4i4e137_app1.pdf]

Multimedia Appendix 2

NHS Peer review 2.

[PDF File (Adobe PDF File), 150KB - resprot_v4i4e137_app2.pdf]

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Abbreviations

CABG: coronary artery bypass graft
CBP: cardiopulmonary bypass
CICU: cardiac intensive care unit
CVP: central venous pressure
DOSA: Day of Surgery Admission
Hct: critical hematocrit
KIM-1: kidney injury molecule-1
LFABP: liver-type fatty acid-binding protein
MMSE: Mini Mental State Examination
NHS: National Health Service
NIRS: near-infrared spectroscopy
RCT: randomized controlled trial
SvO2: mixed venous oxygen saturation

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Protocol

Digital Interventions to Promote Self-Management in Adults With Hypertension: Protocol for Systematic Review and Meta-Analysis

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Abstract

Background: Digital interventions, defined as any intervention accessed and taking input from patients in the form of a computer/Web-based program or mobile phoned-based app, can potentially help empower patients to self-manage long-term conditions such as hypertension. Importantly, digital interventions have the potential to provide patients with personalized information and support for active involvement in treatment as well as cost saving.

Objective: The purpose of this systematic review is to synthesize the evidence for using digital interventions to support patient self-management of hypertension, and determine their impact on control and reduction of blood pressure, other clinical outcomes, quality of life, medication adherence, health service utilization, and economic benefits.

Methods: A systematic search of bibliographic databases including Medline, Embase, CINAHL, and PsycINFO will be undertaken. Abstracts and citations will be independently screened by 2 researchers against predetermined inclusion criteria. Any disagreements will be resolved by discussion and further consideration of the inclusion criteria. Only randomized controlled trials which have been published in peer peer-reviewed journals with a diagnosis of hypertension will be considered. Inclusion criteria will be (1) adults (age ≥ 18 years) with hypertension (as defined by the primary authors); (2) an interactive digital intervention compared with usual care; and (3) outcomes of objectively measured change in blood pressure. Data extraction from identified articles will be undertaken by 2 independent reviewers using a uniform template. The main outcomes are systolic blood pressure (SBP) and diastolic blood pressure (DBP), and quality of life indicators. Secondary outcomes include cost- effectiveness, medication adherence, emotional well-being, and physical activity. Risk of bias of included studies will be assessed using the Cochrane tool.

Results: Our research is currently ongoing. Data will be summarized narratively, and if possible, meta-analyses will be performed to assess the impact of the interventions on outcomes.

Conclusions: By summarizing and synthesizing available data, this review will help inform policy on the use of digital interventions for self-management of hypertension and will clarify areas for further research.

Trial Registration: Prospero 2014: CRD42014010268; http://www.crd.york.ac.uk/PROSPERO/display_record.asp?ID=CRD42014010268 (Archived by WebCite at http://www.webcitation.org/6c5alQQJL)

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KEYWORDS

digital intervention; hypertension; self-management

Introduction

Hypertension has been shown to be the highest attributable risk to death from cardiovascular disease, which is the leading cause of premature mortality worldwide [1]. Reducing levels of blood pressure, even by a small amount, can have a substantial effect on levels of mortality, particularly at the population level [2,3]. However, the rate of control and treatment of hypertension is suboptimal with a large gap found between detection and control [4]. Barriers to adequate blood pressure control include suboptimal treatment by clinicians, suboptimal monitoring due to availability barriers for both patients and clinicians, and suboptimal adherence to medication [5].

The success of blood pressure management depends, to a large degree, on the willingness and ability of the patient to change and maintain certain behaviors and adhere to medication regimens [6]. In England, the National Health Service (NHS) identified self-management as a major priority [7]. Self-management can encompass a wide range of behaviors in addition to medication use and monitoring of symptoms, such as an individual's ability to manage physical, psychosocial, and lifestyle behaviors related to his/her chronic illness and appropriate use of medical care [8]. There is increasing interest in promoting the role of self-management, by which individuals take greater control over their own health and well-being, in supporting the management of long-term conditions such as hypertension [9]. Self-management in hypertension including self-titration and behavioral interventions has been shown to be effective [10-12]. In addition, self-management for hypertension can involve focusing on improving adherence to dietary approaches [13], weight loss [14], increased physical activity [15], smoking cessation [16], and moderation of alcohol intake [17]. A study exploring patients' experiences of an interactive mobile phone-based system designed to support the self-management of hypertension found that it helped them gain an understanding of the interplay between blood pressure and daily life, which resulted in increased motivation to follow treatment [18]. However, few family physicians, by whom most hypertension care is undertaken, have the infrastructure to support such interventions.

One potential method for improving self-management is through the use of interactive digital interventions, which offer the possibility of empowering patients to self-manage their long-term conditions, and by providing patients with better access to personalized information and support for active involvement in treatment, as well as producing significant savings in treatment costs [18-20]. The "interactive" aspect requires contributions from users to produce tailored material and feedback that is personally relevant. Interactive digital interventions are computer-based programs that can combine health information with behavior change, emotional and/or decision support to potentially improve the efficiency of health care by automating routine aspects of patient education, monitoring, and support, while improving services by giving patients convenient 24-hour access to detailed personalized feedback, and allowing health professionals to monitor patient status remotely [21,22]. It has been suggested that well-designed interactive digital interventions can be instrumental in changing patient health-related behavior, improve patient knowledge and confidence for self-management of health, which in turn can result in better health outcomes [11,12]. However, problems with the development and implementation of interactive digital interactions include cost and complexity [23] and high attrition rates (where patients do not use or make suboptimal use of the intervention) [24], respectively. If interactive digital interactions are shown to be an effective adjunct to treatment, further work will be required to address these challenges [25].

Examining the effect of interactive digital interventions in comparison to usual care is important as there is evidence that successful implementation depends on clearly demonstrating their benefits and cost effectiveness to clinicians [26,27]. Self-management interactive digital interventions in a primary care setting offer the opportunity of maximizing both reach and cost savings as the majority of those with hypertension are seen in a primary care setting. Although there are a number of reviews that have examined the impact of self-management in adults with hypertension [28-30], to our knowledge there are none that focus on self-management interactive digital interventions. Moreover, an overview of the literature [31] found 2 Cochrane reviews which concluded that while current evidence offered little support that self-monitoring and mobile phone messaging interventions provided benefit in supporting long-term illnesses, there is a need for further research into these issues [26,32]. Therefore, this systematic review aims to synthesize the evidence for using interactive digital interventions to support patient self-management of hypertension, and determine their impact on control and reduction of blood pressure, other clinical outcomes, quality of life, medication adherence, health service utilization, and health care costs.

Methods

Intervention and Self-Management

The term "digital intervention" can relate to a number of different types of intervention. For the purpose of this review it will include any intervention accessed through a computer (work or home), mobile phone, or other handheld devices, and include a Web-based program, desktop computer program, or apps that provide self-management information. Intervention participants may input information online or offline through the particular device used. The intervention must function without any directive input from health professionals, and be "interactive" in nature. We define "interactive" as requiring contributions from program users (eg, entering personal data and making choices) that alter pathways within the program to produce tailored material and feedback [33]. Studies that only involved sending blood pressure (BP) readings to a remotely

located health professional and receiving advice about medication titration directly from a health professional will be excluded from this review. Interventions that included face-to-face contact and focused on medication adherence will be included if there is also an automated, interactive component without direct health professional mediation (ie, users report SBP interactively then receive automated messages advising them to increase/decrease medication as relevant to their BP levels; trial registration number CRD42014010268).

For the purposes of the review, we define a self-management support intervention as the care taken by individuals toward their own health and well-being comprised by the actions they take (1) to lead a healthy lifestyle, (2) to meet their social, emotional, and psychological needs, (3) to care for their long-term condition, and (4) to prevent further illness or accidents [34].

Eligibility Criteria

Inclusion criteria, based on participants, interventions, comparisons, outcomes, and study design (PICOS acronym) [35] include (1) adult population (aged ≥ 18 years) with hypertension (as defined by the primary authors), (2) an interactive digital intervention (as defined earlier), (3) a comparator of usual care, (4) objectively measured changes in blood pressure (systolic or diastolic), (5) only randomized controlled trials (RCTs) as they present the strongest level of evidence, and (6) only studies published in journals and in English as evidence suggests that limiting studies in this way does not introduce significant bias [36].

Search Methods for Identification of Studies

Searches will be undertaken by a professional systematic review company (York Health Economic Consortium). The search strategy is shown in Multimedia Appendix 1. The databases to be searched are Medline, Embase, CINAHL, PsycINFO, ERIC, Cochrane Library (including CDSR, DARE, Central, and HTA databases), DoPHER and TROPHI (both produced by the EPPI Centre), Social Science Citation Index, and Science Citation Index. These databases will be searched using a combination of subject headings, where available (such as MeSH), and words in the title and abstracts. The resources searched were chosen because they represent a reasonably wide range of core databases covering health care literature and were likely to contain the health care research that is relevant to the review eligibility criteria (RCTs published in peer-reviewed journals excluding literature and conference abstracts). We achieved coverage of journal articles about digital technology through searching the Social Science Citation Index and Science Citation Index.

The search strategy will combine the following concepts and study-type filter: (1) hypertension, (2) digital intervention, (3) self-management, and (4) RCTs.

Search terms for the intervention concept were informed by those used in a previous systematic review conducted on digital asthma self-management interventions [37]. To assess the robustness of the search strategy, PubMed was searched for relevant studies and we identified 10 relevant papers for potential inclusion. We then undertook a hand search of the journals from which the 10 studies were published (Circulation, Journal of American Medical Association [JAMA], American Heart Journal, Journal of Hypertension, Journal of Medical Internet Research, and Journal of Human Hypertension) but no further studies were found. The search strategy was then run to ensure it included the 10 studies among the 5606 papers it identified. The search will also be complemented by contacting experts in the topic under review and by carrying out citation searches for articles citing individual studies that are included in the review [38].

Study Selection

Relevant studies will be ascertained by screening using Distiller software [39] with all identified studies assessed by 2 reviewers. Initially, abstracts will be screened and any potentially relevant studies will be identified and the full-text will be reviewed. Any inter-researcher disagreements over inclusion will be resolved by discussion and a possible third party if a consensus cannot be sought. Excluded studies will be listed with reason(s) for exclusion. The primary outcomes are changes in mean SBP and DBP and quality of life indicators (Table 1).

Table 1. Types of primary outcome measures.

Outcome measure description	Primary outcome	Secondary outcome
Clinical	Mean systolic and diastolic blood pressure	
	Quality of life indicators	
Cognitive		Self-efficacy
Behavioural		Medication adherence
		Dietary change
		Physical activity
		Alcohol intake
Affective		Depression
		Anxiety
		Emotional well-being
		Satisfaction with care
Economic		Health service utilization
		Costs of intervention

Data Extraction and Management

Studies that meet the inclusion criteria will be screened in full by 2 reviewers working independently to extract relevant population, intervention, and outcome data using the Distiller software [39]. Inter-reviewer disagreements will be resolved by seeking consensus or decision by a third party. When papers with duplicate data are found, the largest dataset will be included in any meta-analysis.

Assessment of Quality

Risk of bias will be assessed in each of the included studies by the 2 researchers working independently using the Cochrane collaboration tool for assessing bias [40]. The areas of bias that will be assessed include methods of allocation concealment, generation and presentation of allocation sequence, whether incomplete outcome data were assessed, and whether there was evidence of selective outcome reporting.

Analysis

Details of the populations studied and each intervention will be presented in a table format describing patient and intervention characteristics. We will conduct a narrative synthesis describing, where possible, the components of the interventions including theoretical underpinning, what the mode of delivery was (eg, mobile phone, tablet, personal computer, or Web-based facilitation), how the information was uploaded (online/offline) and where (home/work/other), how ongoing engagement was encouraged, and how often it was used.

Where possible and appropriate we will undertake a meta-analysis that will compare changes between intervention and control participants in outcomes for which adequate data from a minimum of 3 studies are available. We will pool the data for each outcome using mean differences for continuous outcomes and relative risks for dichotomous outcomes. Studies of self-monitoring in hypertension have shown significant heterogeneity and so it is likely that that a random effects model will be required. This decision will be made following estimation of heterogeneity using the I^2 statistic (low <30%; moderate 30-75%; high ≥75%) [41]. Publication bias will be assessed, whenever possible (sufficient number of studies, low heterogeneity), using the Egger regression asymmetry test, the Begg adjusted rank correlation test, and visual examination of funnel plots [42,43]. If high levels of heterogeneity are shown to exist, we will conduct sensitivity analyses if the number of included studies allows, in order to investigate possible sources of heterogeneity including study quality (adequate versus inadequate allocation concealment, low versus high attrition) and sociodemographic factors that could act as effect modifiers (age, gender, and socioeconomic status).

Any subgroup analyses undertaken will be defined a priori. If the data permit, we will undertake the following subgroup analyses: (1) interventions that included self-monitoring of blood pressure versus those that did not, (2) mode of delivery (mobile phone versus other), and (3) primary goal of the intervention (reduction of blood pressure versus any other).

Results

Our research is currently ongoing. Data will be summarized narratively and, if possible, meta-analyses will be performed to assess the impact of intervention on outcomes. The aim is to have all the results completed, written, and published by the beginning of 2016.

Discussion

This review and proposed meta-analysis are part of a study aiming to investigate the best way of providing people with an interactive digital intervention for hypertension that can help them self-manage their health condition, with support as needed from health care professionals. It is thus important to assess previous research on digital interventions to support patient self-management of hypertension and assess the effects, if any, on control and reduction of blood pressure, other clinical outcomes, quality of life, medication adherence, health service

utilization, and health care costs. The results of this review will aid our understanding of current knowledge in relation to the utility of digital self-management interventions for hypertension and identify important research gaps.

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Authors' Contributions

All authors drafted, read, and approved the manuscript.

Conflicts of Interest

None declared.

Multimedia Appendix 1

Search strategy for Ovid Medline(R) in-process and other non-indexed citations and Ovid Medline(R): 1946 to present.

[PDF File (Adobe PDF File), 11KB - resprot_v4i4e133_app1.pdf]

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Abbreviations

BP: blood pressure
DBP: diastolic blood pressure
RCT: randomized controlled trial
SBP: systolic blood pressure

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Original Paper

Human Resource Information Systems in Health Care: Protocol for a Systematic Review

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Abstract

Background: Compared with the eHealth literature as a whole, there has been relatively little published research on the use and impact of information and communication technologies (ICTs) designed to support business functions within health organizations. Human resource information systems (HRISs) have the potential to improve organizational efficiency and effectiveness by facilitating workforce planning, financial and operational administration, staff training, and management analytics. However, the evidence base regarding HRIS in health care is widely distributed across disciplinary boundaries and previous reviews have been somewhat limited in scope. This rigorous systematic review will identify, appraise, and synthesize existing international research on the implementation and impacts of HRIS in health organizations, to provide insights and recommendations that may guide future purchasers, commissioners, implementers, evaluators, and users of such systems.

Objective: The objectives of this review are threefold: (1) to determine the prevalence and scope of existing research and evaluation pertaining to HRIS in health organizations; (2) to analyze, classify, and synthesize existing evidence on the processes and impacts of HRIS development, implementation, and adoption; and (3) to generate recommendations for HRIS research, practice, and policy, with reference to the needs of different stakeholders and communities of practice.

Methods: A high-level scoping review was first undertaken to inform a draft search strategy, which was refined through several cycles of piloting and iteration to optimize its sensitivity and specificity. This was used by the first author, with the help of a medical librarian, to search international electronic databases indexing medical, business, ICT, and multi-disciplinary research. Sources of gray literature and reference lists of included studies were also searched. There were no restrictions on language or publication year. Two reviewers are now screening and coding titles and abstracts for potentially eligible studies, for which full text articles will be retrieved. Reasons for exclusion will be noted for the remaining articles. A structured form will be used to summarize and classify the articles. Any disagreements between reviewers will be resolved through consensus or arbitration by a third reviewer. A PRISMA flow diagram will illustrate the study selection process and ensure transparency of the review. Finally, content experts will be consulted to ensure that important articles have not been missed.

Results: The initial searches have now been completed and the results are being analyzed. The review is expected to be completed and published by the end of 2015.

Conclusions: By synthesizing the existing evidence base, identifying areas in which knowledge is currently lacking, and generating recommendations for research and practice, this review will be a useful resource for decision makers and managers considering or implementing HRIS, as well as encouraging new research in this area.

Trial Registration: PROSPERO International Prospective Register of Systematic Reviews: CRD42015023581; http://www.crd.york.ac.uk/PROSPERO/display_record.asp?ID=CRD42015023581#.VYu1BPlVjDU (Archived by WebCite at http://www.webcitation.org/6ckJCDdCL)

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KEYWORDS

eHealth; health care management; information systems; systematic review; human resource information systems

Introduction

Background

The health sector is complex [1]; while it must inevitably respond to treatment advances, it can be slow to adopt other types of innovation and has lagged behind other sectors in its use of information and communications technology (ICT) [2]. Health organizations are characterized by a dual structure: one for clinical functions and the other for business and support functions [3], which is reflected in their ICT. Despite the importance of business and support systems in health organizations, very little research on their adoption or impacts exists, compared with other areas in health informatics and eHealth [4]. Like clinical information systems, business and support systems vary depending on the department in which they are used (eg, finance, human resources [HR], or procurement) and may be implemented either as standalone systems (eg, integrated procurement system, or inventory management module) or as a part of generic health information or enterprise resource planning (ERP) systems.

This review focuses on one such functional system, human resource information systems (HRISs), either implemented as a standalone system (eg, a payroll module, or as a dedicated multifaceted HR system), or embedded within a broader generic health information system (eg, as an HR module within an ERP system). Although HRIS are vital for the effective operation of health organizations and address many of the information, communication, and training issues of health professionals [2], they are underrepresented within the health, information systems, and management literature. This is despite the fact that "people costs" can account for 65-80% of health organizations' total operating budgets [3] and successful implementation of HRIS in HR departments has been linked to improvements in patient care [2].

The importance of HRIS, and the data that they can generate, has also been highlighted by various global health initiatives [5-15]. For example, the World Health Organization (WHO), in their 2006 World Health report stated that "systems for recording and updating health worker numbers often do not exist, which presents a major obstacle to developing evidence-based policies on human resource development" [16]. Six years later, despite the importance of HRIS for underpinning strong health systems, a 2012 review ([17] cited in [18]), concluded that "universal understanding of the HRIS used in monitoring human resources for health is minimal and baseline information regarding their scope and capability is practically non-existent...[there is a need] for more descriptive research of HRIS globally, including the documentation of impact so as to advance the science and evidence-based practice in this area" [18]. Some researchers have already responded to this request and made their contributions to the international peer-reviewed literature [18,19]. However, because this topic lies at the intersection of informatics, management, and health, most of the existing HRIS studies in health care are spread across several discipline-specific bodies of knowledge, making it difficult to obtain a complete picture of the evidence base.

Formative Scoping

To verify the aforementioned claim, we first searched for and examined existing reviews of HRIS literature, and indeed, as can be seen in Figure 1, these tend to be discipline specific, typically coming either from a business, social science, or ICT perspective.

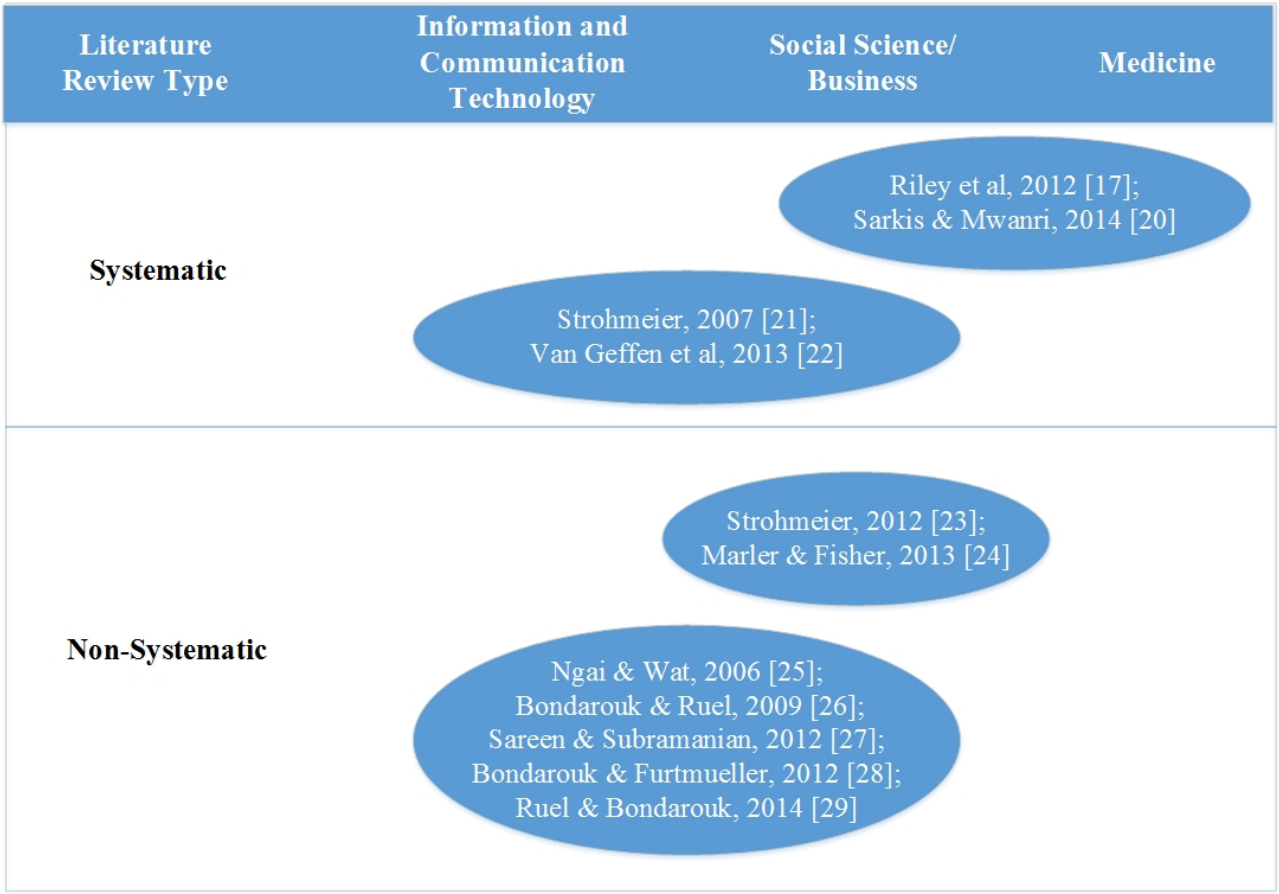
Reviews were classified as systematic if they were either labeled as such, or used a structured search strategy to interrogate named online databases, and clear methods for filtering, summarizing, and synthesizing results. Only a few used a systematic approach [17,20-22], and none encompassed the ICT, social science, business, and health literatures in combination. Only 2 reviews looked specifically at HRIS in health care [17,20], both of which prioritized medical and social science databases, and were limited in scope. The first, published in 2012, was aimed at uncovering baseline information on the use and capability of HRIS in different countries, as a means of understanding the challenges this presents for global health workforce monitoring and development. The second, published in 2013, examined the role of different types of ICTs as a means of enabling continuing professional development to strengthen human resource capacity in health care.

Other nonsystematic literature reviews [23-29] were classified according to their references and the journals in which these references were published. The 6 literature reviews included in Figure 1 [21,22,24,26,28,29] were adopted from "Orchestrating the e-HRM symphony" [30], an inaugural lecture given by Professor T Bondarouk at the University of Twente in December 2014. The other 5 [17, 20, 23, 25, 27] were already known to the authors, based on their background reading.

Our scoping searches, using different keywords, revealed a wide variety of terms and definitions that have been used in the literature to describe information systems aimed at supporting human resource management (HRM) [21], echoing previous scholars' observations on the lack of consistency and agreement in this area [26]. Such systems may be explicitly referred to as *HRIS* (eg, defined as "the composite of databases, computer applications, and hardware and software necessary to collect/record, store, manage, deliver, present, and manipulate data for human resources" [31]), as an *electronic human resource* or *E-HR* system (eg, described as a "created real-time,

information-based, self-service, interactive work environment" [32]) or, more recently, as an *electronic human resource management* or *e-HRM* system (eg, defined as "a way of implementing HR strategies, policies, and practices in organizations through conscious and directed support of and/or with the full use of web-technology-based channels" [33]). These terms are often used to refer to standalone information systems for HR, whereas, in complex organizations, HR functions may be embedded within wider *ERP* systems [34] or *health information systems* [35]. Because the proposed systematic review is designed to be interdisciplinary, comprehensive, and inclusive, it will encompass any study referring to ICT aimed at supporting the administration, management, and development practices of HR in the health sector [36], whether used solely by HR professionals or jointly by HR and health professionals and leaders. This may include such HRM practices as staffing, training and development, compensation and rewards, communication, employee engagement, and performance management [36]. Such systems are not directly related to patient care, although their use in HRM has the potential to indirectly influence care quality and outcomes [37].

Figure 1. Analysis of existing literature reviews on human resource information systems (HRISs).



Aims

We are undertaking an interdisciplinary systematic review to identify and classify existing evidence on development, adoption, implementation, and impacts of HRIS on health organizations. In addition to documenting and classifying the research, we will seek to understand the outcomes and implications of HRIS implementation in health organizations, the theoretical frameworks used to study them, and to identify unexplored areas. This review also aims to support health leaders by summarizing the current evidence base and informing recommendations on HRIS implementation and research.

Objectives

This review concerns the effective development, implementation, and use of ICT platforms and software for supporting the effective management of HR within health organizations and, in some cases, across health care systems. The specific objectives of this review are the following: (1) to determine the prevalence and scope of existing research and evaluation pertaining to HRIS in health organizations; (2) to analyze, classify, and synthesize existing evidence on the processes and impacts of HRIS development, implementation, and adoption; and (3) to generate recommendations for HRIS research, practice, and policy, with reference to the needs of different stakeholders and communities of practice (eg, eHealth, business ICT systems, and HRM).

Methods

Design

This is the protocol for a structured review of scientific and gray literature on HRIS in health care. A PRISMA flow diagram [38] will be followed, to illustrate the study selection process

and ensure transparency of the review (Trial Registration: CRD42015023581).

Data Sources and Search Methods

A high-level scoping review was first undertaken to inform a draft search strategy, which was refined through several cycles of piloting and iteration to optimize its sensitivity and specificity. This was used by the first author, with the help of a medical librarian, to search international electronic databases indexing medical (Cochrane Library, Medline, and EMBASE), Business (ABI/Inform, ASSIA, and Sociological abstracts), ICT (IEEE Xplore), and multidisciplinary (Scopus, Web of Science Core Collection, and ScienceDirect) research, as this topic lies at the intersection of informatics, management, and health. There were no restrictions on language or publication year applied to the literature search.

In addition to the electronic databases, various gray literature sources were also reviewed to identify relevant studies, using a subset of the search terms described in the next section. They included, but were not limited to 4 types of sources. First, WHO reports and working papers were searched using WHO's *Institutional Repository for Information Sharing* and the following query: HRIS OR eHRM OR e-HRM OR "Human resource information system". Second, with respect to professional, marketing, and consulting company reports, the resources/publications/web pages of the following organizations were examined individually to identify relevant articles: Chartered Institute of Personnel and Development, Society for Human Resource Management, Deloitte, Ernst & Young, PricewaterhouseCoopers, KPMG, Towers Watson, McKinsey & Company, Boston Consulting Group, and Sierra-Cedar. Third, white papers and publications of the Healthcare Information and Management Systems Society available at their Resource Library (under "Health IT" topics) were included. Finally, academic theses were identified via Google's search engine using the following query: HRIS OR eHRM OR e-HRM OR "Human resource information system" AND Health AND Thesis. The first 7 pages of the returned results were considered relevant and reviewed. References of qualified articles were "snowballed" to identify other relevant studies. Once data extraction is complete, experts in the field will be consulted to identify any further relevant articles that may have been missed.

Keywords Identification

A comprehensive list of keywords was created for this systematic review [39] through three steps. First, the following types of HR-related terms were identified: (1) HR terminology that the authors were familiar with based on their background readings; (2) terms that were returned when searching under "Human resource" in the US National Library of Medicine's Medical Subject Headings (MESH) browser, such as staff and manpower; and (3) terms articulated within a highly cited expert review analyzing theoretical, methodological, and topical aspects of e-HRM, and in the abstracts of the referenced articles [21] (added during the "Search Query 2" development stage). Second, the ICT-related terms "information systems" and "information technology" were selected based on background readings. Third, general health terminology that the authors were already familiar with were identified: health, health care, hospital, clinic*, and medic*. Health care and care were not sufficiently sensitive. Searching the MESH terms database did not reveal any additional relevant results.

Search Query Development

Three main search queries (Table 1) for the identified keywords were tested (Tables 2-5) by the first author on June 25-26, 2015. Their results were then shared with the review team to jointly identify the search query most likely to ensure the highest level of inclusion of relevant studies.

Table 1. Search query development.

	Search Query 1	Search Query 2	Search Query 3
Search query testing	Table 2	Keywords testing: Table 3[a] Search query testing: Table 4	Table 5
Conclusions	We decided not to proceed with this option as it generated results that were too generic.	Additional keywords [21] were added to the search query. HR terms were combined with ICT terms and inverted commas were used for these combined keywords, so the search would show only relevant results. Insufficiently sensitive keywords that yielded 0 search results in all databases were not included in further searches (Table 3). Insufficiently specific keywords yielding large numbers of irrelevant results were also rejected (Table 3). Keywords yielding more than 5000 results from all databases per keyword are subclassified as "too broad" in Table 3. Total number of keywords included in the search query = 50 The search query was adopted individually for each electronic database. The total number of returns was 18,000, which included studies from various industries. Therefore, the review team jointly decided to limit the search to HRIS studies in health (Search Query 3).	The search query was adopted individually for each electronic database. Total number of returns = 6663

[a]Full keyword testing statistics for each database can be requested by email from the first author.

Table 2. Search query 1 testing.

Search query	Web of Science core collection	Scopus
((HR OR "HR management" OR "HR administration" OR "Human resource" OR "Human resource management" OR "Human resource administration*" OR Workforce OR "Workforce management" OR "Workforce administration" OR Personnel OR "Personnel management" OR "Personnel administration" OR Manpower OR "Manpower management" OR "Manpower administration" OR Employee OR "Employee management" OR "Employee administration" OR Staff OR "Staff management" OR "Staff administration") AND ("Information technolog*" OR "Information system*")) OR ("e-HRM" OR HRIS OR "Electronic Human Resource")	7381	108,256

Table 3. Search query 2 keywords testing.

Status	Keywords
Included	"E-HR" OR "e-HRM" OR eHRM OR HRIS OR "electronic Human resource" OR "HR management system*" OR "HR information system*" OR "HR technolog*" OR "HR management information system*" OR "HR administration system*" OR "HR information technolog*" OR "HR management technolog*" OR "Workforce management system*" OR "Workforce information system*" OR "Workforce technolog*" OR "Manpower management system*" OR "Manpower information system*" OR "Manpower management information system*" OR "Employee management system*" OR "Employee information system*" OR "Employee management information system*" OR "Staff management system*" OR "Staff information system*" OR "Staff management information system*" OR "Staff administration system*" OR "Human resource information technolog*" OR "Human resource management technolog*" OR "Human resource* technolog*" OR "Human resource information system*" OR "Human resource management information system*" OR "Human resource administration system*" OR "Human resource management system*" OR "Personnel information system*" OR "Personnel management information system*" OR "Personnel administration system*" OR "Personnel management system*" OR "Personnel Staffing and Scheduling Information Systems" OR "electronic HRM" OR "Virtual HRM" OR "Web-based HRM" OR "HR Portal" OR "HR Online" OR "HR Intranet" OR "E-recruit*" OR "Electronic recruit*" OR "E-employment" OR "Virtual HR" OR "Web-based HR" OR "Business-to-employee" OR "Employee self service"
Not included (not sufficiently sensitive: no results)	"Human resource administration information system*"; "HR administration information system*"; "Workforce administration information system*"; "Personnel administration information system*"; "Manpower administration information system*"; "Employee administration information system*"; "Employee administration system*"; "Workforce administration system*"; "Personnel administration information system*"; "Manpower administration system*"; "Staff administration information system*"; "HR management information technolog*"; "Human resource administration information technolog*"; "Human resource administration technolog*"
Not included (not sufficiently specific: many irrelevant results)	"HR administration technolog*"; "Workforce management information system*"; "Human resource management information technolog*"; "Personnel technolog*"; "Personnel IT"
Not included (too broad: over 5000 hits for all databases per keyword)	"Personnel IS"; "Electronic learning"; "E-learning"; "Intranet"

Article Screening and Selection

Procedure

Systematic review software (EPPI-Reviewer 4) is used to store, screen, and code all data generated by the search strategy citations. Two reviewers are screening and coding titles and abstracts for potentially eligible studies, for which full-text articles will be retrieved. In cases for which it is impossible to locate the full text of an article, requests will be sent directly to the authors. Reasons for exclusion will be noted for the remaining articles. A structured form will be used to summarize and classify the articles. Disagreements between reviewers will be resolved through consensus or arbitration by a third reviewer. A PRISMA flow diagram will illustrate the study selection process and ensure transparency of the review. Finally, content experts will be consulted to ensure that important articles have not been missed.

Inclusion Criteria

This study has two inclusion criteria. First, any study involving a formal or semiformal approach to the investigation or evaluation of HRIS, whether led by academia, industry (eg, consulting sector), or from within the health care sector will be included. This includes, but is not limited to, studies of HRIS development, implementation, deployment, diffusion, adoption, use, and impacts. Second, studies of broader nonclinical business/administrative/ERP systems that explicitly examine their application to HR practices will also be included.

Participants

There will be no exclusion based on participants/population group.

Interventions

This review is not restricted to intervention studies; however, it will include evaluations of interventions aimed at engaging personnel in the use of HRIS, as well as evaluation studies for which the implementation of HRIS represents an intervention, whether or not this is part of an explicit experimental design.

Table 4. Search query 2 testing.

Database	"E-HR" OR "e-HRM" OR eHRM OR HRIS OR "electronic Human resource" OR "HR management system*" OR "HR information system*" OR "HR technolog*" OR "HR management information system*" OR "HR administration system*" OR "HR information technolog*" OR "HR management technolog*" OR "Workforce management system*" OR "Workforce information system*" OR "Workforce technolog*" OR "Manpower management system*" OR "Manpower information system*" OR "Manpower management information system*" OR "Employee management system*" OR "Employee information system*" OR "Employee management information system*" OR "Staff management system*" OR "Staff information system*" OR "Staff management information system*" OR "Staff administration system*" OR "Human resource information technolog*" OR "Human resource management technolog*" OR "Human resource* technolog*" OR "Human resource information system*" OR "Human resource management information system*" OR "Human resource administration system*" OR "Human resource management system*" OR "Personnel information system*" OR "Personnel management information system*" OR "Personnel administration system*" OR "Personnel management system*" OR "Personnel Staffing and Scheduling Information Systems" OR "electronic HRM" OR "Virtual HRM" OR "Web-based HRM" OR "HR Portal" OR "HR Online" OR "HR Intranet" OR "E-recruit*" OR "Electronic recruit*" OR "E-employment" OR "Virtual HR" OR "Web-based HR" OR "Business-to-employee" OR "Employee self service"
Web of Science core collection	952
Scopus	2482
ScienceDirect[a]	278
ABI/Inform[b]	6771
ASSIA	37
Sociological abstracts	58
Cochrane Library	16
Medline	542
EMBASE	227
IEEE Xplore[c]	1875+4762
Total	18,000

[a]Database was manually set to search in "Abstract, Title, Keywords" to match search strategies in other databases.

[b]Database was manually set to search "Anywhere except full text" to match search strategies in other databases.

[c]Database has limitations on the number of keywords; therefore, the search had to be run several times to ensure that all search query keywords were included.

Comparisons

This is not a review of clinical trials, and we anticipate that most studies will be of the qualitative/investigative type. However, for studies evaluating an intervention, relevant comparators will include baseline measures of efficiency. Examples include payroll processing time, and indicators of impact, such as staff absenteeism, which could theoretically be associated with workload, or patient morbidity, which may be theoretically associated with effective staff deployment. Our inclusion criteria encompass all types of research or evaluation.

Context

Any health organization, including primary, secondary, or tertiary care settings, or health systems where HRIS are implemented at scale may be included.

Exclusion Criteria

Reports that are purely descriptive, only concerned with the computational design aspects of systems, or pure market research will be excluded, as the aim of this project is to glean both softer and harder forms of evidence (eg, social studies, quality improvement projects, and impact evaluations).

Outcomes

Primary Outcomes

Primary outcomes are measures of organizational efficiency, effectiveness, safety, quality, and cost effectiveness.

Secondary Outcomes

Secondary outcomes include the effectiveness of change management processes; indicators of implementation, adoption, and use; perceived benefits and disadvantages; perceived facilitators and barriers; and satisfaction of managers and/or employees.

Table 5. Search query 3 testing.

Database	("E-HR" OR "e-HRM" OR eHRM OR HRIS OR "electronic Human resource" OR "HR management system*" OR "HR information system*" OR "HR technolog*" OR "HR management information system*" OR "HR administration system*" OR "HR information technolog*" OR "HR management technolog*" OR "Workforce management system*" OR "Workforce information system*" OR "Workforce technolog*" OR "Manpower management system*" OR "Manpower information system*" OR "Manpower management information system*" OR "Employee management system*" OR "Employee information system*" OR "Employee management information system*" OR "Staff management system*" OR "Staff information system*" OR "Staff management information system*" OR "Staff administration system*" OR "Human resource information technolog*" OR "Human resource management technolog*" OR "Human resource* technolog*" OR "Human resource information system*" OR "Human resource management information system*" OR "Human resource administration system*" OR "Human resource management system*" OR "Personnel information system*" OR "Personnel management information system*" OR "Personnel administration system*" OR "Personnel management system*" OR "Personnel Staffing and Scheduling Information Systems" OR "electronic HRM" OR "Virtual HRM" OR "Web-based HRM" OR "HR Portal" OR "HR Online" OR "HR Intranet" OR "E-recruit*" OR "Electronic recruit*" OR "E-employment" OR "Virtual HR" OR "Web-based HR" OR "Business-to-employee" OR "Employee self service") AND (Health OR Healthcare OR Hospital* OR Clinic* OR Medic*)
Web of Science core collection	105
Scopus	838
ScienceDirect[a]	86
ABI/Inform[b]	731
ASSIA	20
Sociological abstracts	10
Cochrane Library	16
Medline	428
EMBASE	134
IEEE Xplore[c]	4295
Total	6663

[a]Database was manually set to search in "Abstract, Title, Keywords" to match search strategies in other databases.

[b]Database was manually set to search "Anywhere except full text" to match search strategies in other databases.

[c]Database has limitations on the number of keywords; therefore, the search had to be run several times to ensure that all search query keywords were included. Original number of returns was 4847. This result was then checked and cleaned of duplicates with the help of systematic review software (EPPI-Reviewer 4). In total, 552 duplicates were removed during this process.

Data Abstraction

The following information will be extracted from each of the eligible studies: authors/institutional affiliation; year; setting (type of organization, country or region in which the study was conducted); innovation stage (eg, design, piloting, implementation); journal discipline; HRIS function/activity; research purpose/questions; theoretical basis (if specified, or if this can be deduced from the author's description); study design; main findings; and conclusions. Other fields may be added as the analysis progresses.

Data Analysis and Synthesis

It is expected that data synthesis will be descriptive, only due to the interdisciplinary nature of the review, variety of study designs, outcomes, and heterogeneity of technology functions and their applications. The data, therefore, will be descriptively summarized and narratively reported.

Critical Appraisal Techniques

Two reviewers will independently assess included studies to eliminate the risk of selection bias; the advice of the third reviewer will be sought in cases of disagreement. We anticipate that most studies will be qualitative. In line with other systematic literature reviews of qualitative research [40,41], we will use the Critical Appraisal Skills Programme (CASP) qualitative checklist [42] to assess study quality.

Results

The initial searches have now been completed and the results are being analyzed. The review is expected to be complete and published by the end of 2015.

Discussion

This review aims to present an unbiased summary and analysis of the existing evidence on HRIS development, adoption, implementation, and impacts regarding health organizations worldwide. We believe that the review will provide a useful resource for decision makers and managers considering or already implementing HRIS. We also expect that the recommended directions for future research that will follow from this review will generate more discussion and research among scholars with interest not only in HRIS, but also in business and support systems in health care.

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Authors' Contributions

AT developed the protocol and search strategy, with input from the other authors. AT and RB will conduct the screening and apply the inclusion/exclusion criteria, with third-party arbitration by CP, when necessary. Included articles will be subjected to data extraction and synthesis by AT, with verification by the entire team.

Conflicts of Interest

None declared.

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Abbreviations

E-HR: electronic human resource
E-HRM: electronic human resource management
ERP: enterprise resource planning
HR: human resource
HRIS: human resource information system
HRM: human resource management
ICT: information and communication technology
MESH: Medical Subject Headings
WHO: World Health Organization

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Original Paper

Western Australian Public Opinions of a Minimum Pricing Policy for Alcohol: Study Protocol

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Abstract

Background: Excessive alcohol consumption has significant adverse economic, social, and health outcomes. Recent estimates suggest that the annual economic costs of alcohol in Australia are up to AUD $36 billion. Policies influencing price have been demonstrated to be very effective in reducing alcohol consumption and alcohol-related harms. Interest in minimum pricing has gained traction in recent years. However, there has been little research investigating the level of support for the public interest case of minimum pricing in Australia.

Objective: This article describes protocol for a study exploring Western Australian (WA) public knowledge, understanding, and reaction to a proposed minimum price policy per standard drink.

Methods: The study will employ a qualitative methodological design. Participants will be recruited from a wide variety of backgrounds, including ethnic minorities, blue and white collar workers, unemployed, students, and elderly/retired populations to participate in focus groups. Focus group participants will be asked about their knowledge of, and initial reactions to, the proposed policy and encouraged to discuss how such a proposal may affect their own alcohol use and alcohol consumption at the population level. Participants will also be asked to discuss potential avenues for increasing acceptability of the policy. The focus groups will adopt a semi-structured, open-ended approach guided by a question schedule. The schedule will be based on feedback from pilot samples, previous research, and a steering group comprising experts in alcohol policy and pricing.

Results: The study is expected to take approximately 14 months to complete.

Conclusions: The findings will be of considerable interest and relevance to government officials, policy makers, researchers, advocacy groups, alcohol retail and licensed establishments and organizations, city and town planners, police, and other stakeholder organizations.

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KEYWORDS

alcohol; addiction; policy; minimum pricing

Introduction

Background

Excessive alcohol consumption has a direct negative impact on economic, social, and health outcomes. Regular moderate-to-heavy alcohol exposure is associated with numerous chronic health conditions, including liver cirrhosis, a range of cancers, and mental health problems [1-3]. Treating chronic harm from alcohol also places considerable burden on health care services [4]. In addition, acute patterns of alcohol consumption such as risky single-occasion alcohol consumption (ie, "binge" drinking) are associated with harmful outcomes such as drunk driving, violence, social disorder, and criminal behavior [5]. Binge drinking also has serious financial implications, including substantive costs for emergency services, such as ambulances or police services attending incidents caused directly or indirectly as a result of excessive alcohol consumption.

In Australia, studies by Collins and Lapsley [6] and the Foundation for Alcohol Research and Education have estimated the annual social and economic cost of alcohol consumption to be between AU $15 billion and AU $36 billion. Reducing the harmful outcomes associated with excess alcohol consumption is a frequently discussed component of Australian federal and state government policy agendas [4,7-10]. Policies influencing price are most effective in reducing population level consumption [11-14]. As a complementary policy to taxation, minimum pricing has received increased national and international attention in recent years [7,8,10,15,16]. Minimum pricing has attracted public health interest because it raises the cost of alcoholic beverages in proportion to their strength or alcohol content and, accordingly, targets beverages with high alcohol content sold at very low prices. While the policy has been implemented in a small number of countries to date, available empirical evidence indicates that population-level drinking is significantly reduced by minimum pricing [16]. However, evidence alone is not sufficient to ensure successful uptake of policy; public opinion is a key potential barrier to implementation [9].

Against this background, investigating attitudes to minimum pricing in Australia is an important research priority for public health advocates interested in policy avenues to reduce excessive consumption. An understanding of the Australian public's attitudes and beliefs toward minimum pricing will provide critical insights into the likelihood of acceptability or opposition and inform public information campaigns that may pave the way for its introduction. The aim of this qualitative study is to investigate public beliefs and attitudes toward the introduction of a minimum price per standard drink policy. The study will be the first to investigate perceptions regarding alcohol minimum pricing in Australia and it will not only seek to provide evidence as to whether the public will support the introduction of minimum pricing, but also perceptions as to what circumstances or conditions may maximize its acceptability.

Minimum Price Policies

A panoply of alcohol control policies have been proposed and implemented worldwide to reduce excessive alcohol consumption (see Babor et al for review [12]). Policies focusing on the price of alcohol have been found to be most effective in reducing excessive alcohol consumption [11,12,14]. Research has shown that population-level alcohol consumption is inversely related to the price of alcoholic beverages [8,17]. While alcohol duty and taxation have increased, so have average incomes such that alcohol, by comparison, has become more affordable. Alongside the increased affordability of alcohol, approximately 80% of the Australian population aged 14 and over report some level of alcohol consumption [4]. While annual rises in duty have reduced alcohol consumption, there is still considerable scope for consumers to access heavily-discounted alcohol. For example, consumers can substitute and alter their drinking habits to avoid higher taxes (eg, by switching to alternative, cheaper beverages such as cider [9]). Minimum pricing involves setting a "floor" or minimum price per standard drink, below which it would be illegal to sell alcohol. Unlike taxation, this policy cannot be circumvented by deep-discounting, below-cost strategies or promotions (ie, "buy-one-get-one-free," "2-for-1," or "multibuy" offers [18]). Modeling studies have indicated that minimum pricing would be effective in reducing excessive alcohol consumption and binge drinking [10,11,14,15,19-21]. Forms of minimum price policies have been implemented in a small number of countries such as Ukraine, Uzbekistan, Russia, the Republic of Moldova, provinces in Canada, and some US states (eg, Connecticut) [7,9,16,17,19].

While several leading public health organizations (eg, the National Alliance for Action on Alcohol, which represents 75 organizations) and organizations concerned with alcohol consumption in specific areas have voiced support for minimum pricing, public opinion on the policy is unclear. In fact, there is a dearth of studies worldwide investigating public opinion to minimum pricing [11,15,19,21]. Assessing public opinion and response to public health policies based on legislation, such as alcohol pricing policies, plays an important part in policy development. Government officials may be reluctant to implement policies that are unpopular or poorly understood by the public because of perceived fear that it might adversely impact them at the polls or that they might lose support of commercial interests [9].

Few studies have investigated public opinion toward minimum pricing [15]. An initial study in the United Kingdom revealed that responses of members of the general public to minimum pricing were "lukewarm" and "less than enthusiastic." Participants indicated that they thought the policy would be ineffective and disliked. Participants also stated concern and skepticism regarding the aims and structure of minimum pricing, requesting greater transparency regarding where the additional revenue generated would be directed. Participants indicated that they would be more positively inclined toward the policy if the revenue generated was hypothecated to alcohol harm prevention and treatment strategies.

Study Protocol

This protocol outlines the design of a study examining the knowledge, attitudes, and beliefs of members of the Western Australian (WA) public regarding a minimum price per standard

alcoholic drink policy. The study will adopt a qualitative design to generate participant-led data on minimum pricing, including basic awareness, knowledge, and understanding of the policy and attitudes and beliefs toward its effect and possible introduction.

Methods

Design and Procedure

The study will employ a qualitative design, involving focus groups to gain in-depth and detailed insight into the awareness and knowledge of minimum pricing in members of the WA general public, their attitudes and beliefs toward the policy, and suggestions for increasing acceptability if the policy was introduced in WA. The study will be conducted over a 14-month period and recruit 10-15 focus groups comprising 8-10 adults in each group. Focus groups will last approximately 1 hour, led by a trained facilitator, and follow a semistructured standardized question schedule (Table 1) to ensure consistency and facilitate comparison and analyses.

The focus group schedule will be separated into 3 main parts. First, participants will be asked to indicate their understanding of the phrase "minimum pricing policy" with respect to alcohol. After assessing the participants' knowledge, the facilitator will then provide a clear-language explanation of the policy for all participants. Information given by the facilitator will include a clear outline of the proposal, as well as previous evidence and findings related to the proposal. Second, the facilitator will subsequently enquire about participants' attitudes and beliefs toward minimum pricing. Third, focus group participants will be asked to consider ways in which the policy could be made more acceptable and effective.

In-depth discussion will be stimulated by the facilitator throughout each part of the discussion. Data will be recorded on 2 voice-recording machines placed strategically to capture all voices in the room. The facilitator will encourage participation in an autonomy-supportive manner and prompt participants to be candid in their views and freely elaborate on their responses. Visual aids will also be introduced and explained to participants to assist with their understanding of how the minimum price policy will affect the price of alcoholic beverages.

Participants

Eligible participants will include WA adults from a diverse cross section of backgrounds, including students, blue/white collar workers, minority groups, unemployed, and retired workers. Individuals younger than 18 years of age will be ineligible to participate as they are not old enough to purchase alcohol in WA. Further, participants who are considered harmful drinkers, according to a screening tool administered prior to the beginning of the focus group (eg, Fast Alcohol Screening Test), will be excluded. Any harmful drinkers identified during the course of the recruitment phase will be referred to alcohol awareness and counseling services.

Participant Recruitment

Participants will be recruited through targeted advertisements and the research team's existing collaborative links with the community, including schools, local employers, local clubs and organizations, and job seeker's pages in local newspapers and in Perth job centers. Advertising materials (eg, posters and emails) will be developed to inform potential participants of the study aims and encourage them to contact the primary researcher to join a focus group. Posters advertising the study will be disseminated across the Perth metropolitan area, as well as emails sent to a wider catchment area in the neighboring suburbs of Perth. Email addresses will be sourced through word-of-mouth and Internet sites of groups and clubs.

One focus group will be exclusively female as research suggests women are likely to hold particular views and beliefs regarding alcohol drinking [22]. An additional 3-4 focus groups will target young professionals recruited from companies that employ white and blue collar male and female workers, groups that have reported high levels of alcohol consumption [23]. We plan to conduct approximately 4 focus groups among older adults from different ends of the socioeconomic spectrum. We will also plan to conduct focus groups in a sample of unemployed people. Finally, we aim to conduct 2 focus groups comprising people from the most populous ethnic minority groups in Perth, namely people from Chinese (eg, 2.9% of the Perth population) and South Asian (eg, Indian, Pakistani, Bangladeshi; 0.8% of the Perth population) backgrounds or have these groups represented in the sample. These focus groups will reflect a diversity of views, attitudes, and views.

Table 1. Interview questions for minimum price policy focus groups.

Focus group topic	Key questions	Follow-up questions
Reaction to minimum pricing	What are you your immediate thoughts about the minimum price policy?	What do think about the idea of minimum pricing?
		Do you think it is a good idea? Do you think it will work?
		Are you in favor of it?
	What concerns, if any, would you have about minimum pricing policy of alcohol?	
	What information/conditions would you like about the minimum pricing policy of alcohol before it was introduced?	
	What do you think are the possible outcomes of a minimum pricing policy?	
	Do you think that introducing minimum pricing policy will actually reduce how much people drink?	
	Who do you think will be most influenced by price increases?	
	How do you think price increases might influence your drinking?	Would minimum pricing policy change how much or what you drink?
	Do you think a minimum pricing policy will reduce alcohol-related harm, crime, social disorder?	
	Is alcohol different to other commodities? Would you continue to drink excessively regardless of any price increases?	
	Do you think minimum pricing policy will affect poor and rich people differently?	
	What impact do you think minimum pricing policy of alcohol will have on underage drinking? Heavy drinkers?	
Would you support the introduction of a minimum pricing policy of alcohol?	What are your reasons for supporting the policy? Or not?	
	What are the possible advantages of introducing the policy?	
	What possible negative effects do you think the policy may have?	
	Do you think the policy is aimed at particular subgroups of the Australian population? What are your reasons for this?	
	Do you think the policy fairly or unfairly focuses on certain subgroups in society?	
	Do you think the policy will work?	
What factors do you think would make a minimum pricing policy more tolerable or accepted by Australians?	What do you think would make the policy more effective?	
	Are there any additional steps (eg, information, public education) you think the government could take to help make this policy more acceptable?	

Data Analytic Method and Sample Size

Given the paucity of research regarding public perceptions about minimum pricing, the adoption of a qualitative approach using focus groups is appropriate and fits well with the general aim to provide a comprehensive overview of people's knowledge, attitudes, and beliefs of minimum pricing and to construct a

model of the factors affecting the acceptability of the policy. Our qualitative approach will be data-driven following recommendations in the literature by qualitative research methodologists [24,25].

Although the approach is not guided by theory, it is not atheoretical. Instead, we will use a theory-building, inductive approach rather than a traditional theory-testing, deductive approach. Specifically, data in the form of transcripts of focus group discussions on minimum pricing will be subjected to inductive thematic content analysis [24] for relevant themes that will not only give important information about people's knowledge of the policy but also provide detail on the relationship between key factors regarding minimum pricing and its acceptability and effect on drinking behavior. Our approach assumes no predetermined categories as it is important that themes emerge from the data during analysis with the focus on providing an in-depth understanding of the themes rather than "smoothed down" generalizations.

Based on Lonsdale et al [15], 10-15 focus groups will be recruited, giving an approximate sample size of between 80 and 150 participants. Once 10 focus groups have been completed, a preliminary data analysis using inductive thematic content analysis will be conducted. At this stage, if no new themes appear to be gained through further data collection, data saturation will be deemed to have occurred and the data collection stage of the study will end. Data analysis and judgment on data saturation will be verified by 2 experienced researchers in qualitative analyses.

Measures

A research question protocol will be developed (Table 1) to ensure every focus group is asked the same questions pertinent to the research aims. This protocol will be developed based on the advice of a project steering group, comprising experts on alcohol policy, researchers on alcohol behavior, and stakeholders. It will then be pilot tested on several members (at least 10) of the general public to ensure that there are no ambiguities or errors and that the transition between topics allows for proper in-depth exploration of the key issues.

Results

The study is expected to take approximately 14 months to complete. This includes 2-3 months for the development of materials (eg, question protocol, study posters). Approximately 1-2 months will be required for advertising and recruitment, followed by 4-5 months of data collection. It is then expected to take between 3 and 4 months to transcribe the data, analyze and agree on emergent themes, and write the results into a coherent framework.

Discussion

Expected Outcomes

This study protocol outlines study methodology to investigate attitudes and beliefs toward minimum pricing and identify avenues for increasing acceptability in members of the WA general public. This study will be the first of its kind in Australia and add to a nascent literature exploring public opinion about minimum pricing [15]. Given that there is increasing support for the introduction of minimum pricing around the world—particularly in Canada, the United Kingdom, and Ireland—this study is especially timely. Findings will likely be of considerable interest to researchers, policy makers, and stakeholders interested in managing and curbing excessive alcohol consumption. Further, the methodology could be used by other research and policy groups interested in examining public opinion to minimum pricing.

Although the planned analysis will be inductive and focus on emergent themes generated from the data, such a process does not occur in a "vacuum" independent of other literature and previous research. Based on previous research we can therefore form a candidate list of themes that may emerge from the data [23]. This will be used as a starting point for comparisons but will not be the sole focus of the analysis. The following key themes are expected to emerge in terms of beliefs regarding the minimum pricing alcohol policy: improving health, promoting law and order, saving public money, violation of personal freedoms, and indirect taxation. We also anticipate that knowledge and understanding of the policy, how it differs from other alcohol pricing policies, and its effects on differing levels and patterns of alcohol consumption in Australia may also differ from research conducted in countries with a history of publicity and debate over the introduction of minimum pricing, such as the United Kingdom [15]. The lack of knowledge and information regarding the policy may mean that understanding that it would have negligible effect on moderate drinkers' expenditure on alcohol [26] and, hence, support for the policy, may be compromised in this sample. In addition, ways of making the policy more acceptable will involve providing information about health, economic, and social benefits of the policy, such as illustrating how the policy might save money by reducing the economic burden on health service, providing information about the risks of drinking and current problems, giving practical advice on how the pricing might affect "within-limit" alcohol drinkers, and framing the changes as socially responsible [15].

Strengths and Limitations

The unique perspective and in-depth exploratory qualitative approach are major strengths of the current proposed study. It is the first study to examine the public interest case for minimum pricing in Australia and will provide valuable information on the perceptions of the general public that may assist in the development of legislation and messages toward the introduction of the policy. Furthermore, the methodological approach facilitates in-depth exploration of participants' views and beliefs, and the use of a predetermined question protocol enables group comparisons.

There are some limitations with the current research. First, the researcher will not blind the study or background research. It may be the case that participants in each focus group ask the researcher for his or her views of minimum pricing. In this situation, the researcher will be briefed to remain neutral, will remind the group that the goal of the research is to canvas opinion and redirect the discussion to eliciting participants'

views and beliefs. Having an informed researcher is also an advantage in the context of the current study as confusion has been found to surround minimum pricing [15]. Second, due to logistical and financial considerations, this study will focus on a sample of adults from WA. Coupled with the small sample, this means that any extrapolation of findings may not generalize to the wider Australian public.

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Authors' Contributions

MSH conceived the study and wrote the original protocol. DAK and MSH drafted the manuscript and contributed to the protocol design. NC, TC, and MD assisted in developing the protocol and read and approved the final manuscript. SH helped to draft the manuscript and provide expertise in relation to the proposed data collection and qualitative analysis.

Conflicts of Interest

None declared.

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Abbreviations

WA: Western Australia

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Original Paper

Understanding Psychosocial and High-Risk Sexual Behaviors Among Detained Juveniles: A Descriptive Study Protocol

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Abstract

Background: African American women are disproportionately impacted by sexually transmitted infections (STIs), such as chlamydia and gonorrhea, which are known risk factors for human immunodeficiency virus (HIV) infection. STIs, particularly chlamydia and gonorrhea, are even more prevalent among young African American women with a juvenile detention history. The population with experiences with the criminal justice system has greater rates of STIs and is diagnosed more often with mental health issues, often related to sexual abuse or intimate partner violence, compared to peers who have not been detained by law enforcement. Psychosocial factors, especially those related to intimate relationships (ie, the imperativeness of being in a relationship and the power one has in their relationship), have emerged as important explanatory factors for acquiring STIs, including HIV, and a component of risk reduction interventions.

Objective: To investigate more comprehensively the relationship between psychosocial risk factors and STIs, including HIV, as it relates to reduction and prevention of these diseases. The long-term goal is to improve the effectiveness of evidence-based interventions with a major focus on intimate relationship dynamics.

Methods: This descriptive study surveys young women (ages 13-17) who have been detained (incarcerated) by a department of juvenile justice. In addition to being female and detained, eligibility criteria include being detained longer than 30 days and being free of cognitive impairments. This study will include young women from one juvenile detention center. The primary outcomes to be measured are STI knowledge, intimate relationship dynamics (ie, imperativeness and power), and high-risk sexual behaviors. High-risk sexual behaviors will be assessed using data extracted from health records.

Results: Preliminarily, we have received assent from 26 primarily young African American women. The majority of participants (81%) had inadequate knowledge about STIs, 52% perceived a lack of power in their relationship, 56% were fearful of negotiating condom use, and 60% were not comfortable refusing sex. Interestingly, a majority of participants (68%) did not perceive a relationship as imperative.

Conclusions: When enrollment and data collection are completed, it is expected that the primary outcome of intimate relationship dynamics (ie, imperativeness and power) will be associated with high-risk sexual behaviors and having an STI. Further, the

findings are expected to provide guidance in developing a risk reduction intervention, for the population in which psychosocial factors related to intimate relationships will be central.

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KEYWORDS

adolescent; African Americans; HIV infections; mental health; risk factors; risk reduction behavior; sexual behavior; sexually transmitted diseases; spouse abuse; unsafe sex

Introduction

The National HIV/AIDS Strategy for the United States has made risk reduction, prevention, and treatment a high priority for populations most affected by and those at great risk for acquiring the disease [1]. African American women represent 63.5% of new human immunodeficiency virus (HIV) infections across all racial and ethnic female populations, with the rate of infection for this group estimated to be 228.8 per 100,000; this is among the highest in the southeast [2-5]. Further, African American women are disproportionately impacted by sexually transmitted infections (STIs), such as chlamydia and gonorrhea, which are known risk factors for HIV infection [6-10].

Understanding the relationship between HIV and other STIs, such as chlamydia, gonorrhea, and herpes simplex virus (HSV), is a component of the National HIV/AIDS Strategy for the United States [1], particularly with respect to the effort to reduce and prevent the disease. This study aims to investigate more comprehensively the relationship between psychosocial risk factors and STIs, including HIV, as it relates to reduction and prevention of these diseases. STIs are a significant health issue among young adults (≥ 20 years old) in Georgia and across the nation [6,11,12]. In 2014, the Centers for Disease Control and Prevention (CDC) reported that STIs were particularly problematic in Georgia and in the south in general [6]. Georgia ranked ninth in the United States for chlamydia, eighth for gonorrhea, and first for primary and secondary syphilis [6].

African American females (adolescents and adults) are at greater risk of acquiring chlamydia and gonorrhea than any other racial or ethnic female population [2,13-17]. STIs, particularly chlamydia and gonorrhea, are even more prevalent among young African American women with a juvenile detention history [10]. Despite the disproportionate rate of STIs among young African American women with a juvenile detention history and their risk for HIV, only a few studies related to these diseases have included this population [10]. We examined a similar population (young African American women who are vulnerable but not detained) to understand risk factors related to STIs/HIV and our population.

Vulnerable young African American women without a history of incarceration share similar characteristics with young African American women with a juvenile detention history, such as limited educational achievement, low socioeconomic status, limited utilization of health care services, unprotected sex, and substance use [18,19]. Studies of vulnerable young African American women suggest that these individuals exhibit high-risk sexual behaviors compared to nonvulnerable peers, have more lifetime sexual partners, engage in sex while high on drugs and alcohol, have low condom self-efficacy, do not use condoms consistently, and are more likely to have STIs [15,18,20].

Despite similarities between young African American women with and without juvenile detention history, the population with criminal justice system experiences has a greater rate of STIs and is diagnosed more often with mental health issues, often related to sexual abuse or intimate partner violence (IPV), compared to peers who have not been detained by law enforcement [10,18,19]. Studies investigating trauma (ie, sexual abuse and IPV) among populations who do not have a detention history have found a strong association between psychosocial factors (eg, intimate relationship dynamics), incidence of STIs, and risk of HIV [16,17,21].

Psychosocial factors, especially those related to intimate relationships (ie, the imperativeness of being in a relationship and the power one has in a relationship), have emerged as an important explanatory factor in the acquisition of STIs, including HIV, and component of risk reduction interventions [16,17,22-24]. As psychosocial and explanatory factors for STI/HIV risk, intimate relationships require much further investigation, given the history of physical abuse, sexual exploitation, and mental health issues experienced by many young women, and particularly young African American women, with a juvenile detention history.

Methods

Design

This descriptive study surveys young women (ages 13-17) who have been detained (incarcerated) by a department of juvenile justice (DJJ). This study aims to understand the association between STIs/HIV and psychosocial factors (intimate relationship dynamics) in a juvenile justice population, which has higher rates of mental health issues, substance use disorders, and trauma (eg, sexual abuse and exposure to violence) than the population without a criminal justice history. Further, this study aims to identify explanatory risk factors related to STIs. The long-term goal is to improve the effectiveness of evidence-based interventions (EBIs) that have a major focus on intimate relationship dynamics.

Participants

This study will include young women from one juvenile detention center. The DJJ where the study will take place has three primary levels of supervision: community (probation and parole), regional youth detention centers (short-term detention), and youth development campuses (long-term commitment). The population for this study will come from a regional youth detention center (RYDC). We identified the population at the

RYDC as appropriate for our investigation into STIs and psychosocial factors, since this population likely will have had more recent experiences outside detainment and psychosocial issues (eg, intimate relationships) may be more salient compared to peers residing at a youth developmental campus (YDC).

Young women projected to have commitments less than 30 days will be excluded, since stays of this duration likely will not allow sufficient time to recruit parents/guardians via mail and during on-site visitations, which occur two days per week (Tuesdays and Saturdays). Further, the population undergoes physical, mental/behavioral, dental, and security assessments upon entry into a facility; these assessments require several days, and during this time study personnel will not have access to the population. The inclusion criteria are listed in Table 1.

Table 1. Inclusion and exclusion criteria.

Characteristic/Factor	Inclusion	Exclusion
Facility	Regional Youth Detention Center	All other facilities
Race	All	None
Gender	Female	Male
Age	≥ 13	≤ 12
Projected detention	≥ 30 days	< 30 days
Cognitive capacity	Nonimpaired	Impaired
Reoffense	Allowable	Not applicable

Recruitment

Investigators will collaborate with facility staff to identify young women who meet the inclusion requirements. Once a list of eligible participants is created, parents/guardians of minors or nonemancipated youths will be recruited via mail and during on-site visitations.

After recruitment of parents/guardians, investigators will recruit adolescents. Participants who meet the inclusion criteria will be recruited for one session; interactions with the young women will require approximately 15 minutes. In addition to being detention centers, RYDCs function as schools, providing a similar structure that would be found in a school district outside of the juvenile justice system. Thus, recruitment will occur weekly at the end of a health education class, where potential participants will be invited to participate in a brief paper survey that will require approximately 5 minutes to complete. The survey will be administered face-to-face by study personnel during the health education class.

Attrition will be measured using an enrollment log that will list participants who assent/refuse to participate, parents/guardians who consent, youths who withdraw their assent, and the population that is released earlier than anticipated. The enrollment log will allow study personnel to identify potential reasons why participants were lost from the study (eg, early release).

Assent and Informed Consent

All participants will be required to sign an IRB approved assent form (≤ 17 years old) or consent form (18 and older). Consent of parents/guardians will be required for youths 17 years old and younger. Parents/guardians will be mailed a consent form with a self-addressed and stamped envelope to return their document to the lead investigator. Investigators will follow up in one week with parents/guardians who have not responded. Consent of parents/guardians also will be sought during visitations with their child/children.

Once parental/guardian consent has been obtained, assent will be sought from potential participants who are minors. To minimize embarrassment for adolescents whose parents/guardians do not consent to their participation and to limit peer pressure, all participants will be provided a research packet (two assent/consent forms and one survey) during a health education class. Participants will retain a copy of the assent/consent form and return one to the investigator. Investigators will explain to potential participants what the study entails, what is expected to be learned, why it is important, what participants' contribution will be, their right to participate or not, and the fact that participation will have no impact on parole or leniency, as mandated by the Code of Federal Regulation, U.S. Department of Health & Human Services, Protection of Human Subjects (45 CFR 46) [25].

Hypotheses

This study hypothesizes that young women (13 and older) who have low STI knowledge and relationship power, but high relationship need, will have greater odds of reporting high-risk sexual behaviors, having a mental or behavioral health issue, and being diagnosed or treated for at least one STI (Hypothesis 1). We also hypothesize that there will be race and ethnic disparities for STIs (Hypothesis 2).

Outcome Measures

The primary outcomes to be measured are knowledge, intimate relationship dynamics (ie, imperativeness and power), and high-risk sexual behaviors. Knowledge about STIs will be measured using the STD-Knowledge Questionnaire (STD-KQ), a 27-item instrument with a Cronbach alpha of .86 and test-retest reliability of .88 [26]. Intimate relationship dynamics will be measured using an instrument developed at Emory University's Center for AIDS Research. The instrument is comprised of 27 items, which assess relationship imperativeness and three psychosocial factors (ie, relationship power, self-efficacy to refuse sex, and fear of abuse) [21]. The subscales of relationship power, self-efficacy to refuse sex, and fear of abuse have

Cronbach alphas of .70, .87, and .89, respectively [21]. High-risk sexual behaviors will be assessed using the following data from participants' health records: history of substance use, history as a commercial sex worker, placement in foster care, homelessness, history of having a mental/behavioral health diagnosis, and history of having more than one sexual partner at a time.

Planned Analyses

Statistical analyses will be conducted using SAS 9.4. Frequencies and proportions will be used to describe all discrete data. Means, medians, and standard deviations will be used to describe continuous data. Logistical regression models will be used to identify significant psychosocial factors (relationship imperativeness, relationship power, perceived self-efficacy to refuse sex, and fear of abuse), high-risk sexual behaviors, STI knowledge, and demographics (eg, race, ethnicity, and age). Odds ratios and 95% confidence intervals also will be reported. The chi-square (χ^2) statistic or Fisher's exact test will be employed to determine whether or not the rate of STIs are different across populations.

Ethics

This protocol has been approved by the Institutional Review Board at Georgia Regents University (protocol number 631921-4) and by the Research Review Committee of the Georgia Department of Juvenile Justice.

Results

This study was launched in February 2015 and is actively recruiting participants. Preliminarily, 26 out of 37 young women assented to participate, reflecting a 70% assent rate. We have not asked participants to give reasons for their refusal to participate, since the survey is administered in a classroom setting, and sharing these reasons may result in participant discomfort or their perception of the question as a form of coercion. Most participants are African American and their mean age is 15.75 years (SD 1.22) (see Table 2).

Table 2. Preliminary demographics.

	Age					
	13	14	15	16	17	Total
	n (%)					
African American	2 (8)	2 (8)	2 (8)	5 (21)	5 (21)	16 (67)
Hispanic/Latina	0 (0)	0 (0)	3 (13)	1 (4)	0 (0)	4 (17)
White	0 (0)	0 (0)	2 (8)	1 (4)	1 (4)	4 (17)
Total	2 (8)	2 (8)	7 (29)	7 (29)	6 (25)	24[a]

[a] Missing data = 2

Preliminary data for the knowledge outcome indicate that 81% (21/26) of participants responded incorrectly to at least 7 out of 12 items regarding STIs. A median split, as used by Raiford, Seth, and DiClemente in a related study [21], defined high and low responses for power, fear of abuse, and sex refusal. A majority of participants, 52% (13/25), perceived they did not have power in their relationships, 56% (14/25) feared negotiating condom use with partners, and 60% (15/25) perceived a lack of self-efficacy to refuse sex. However, emerging data also indicate that 68% (17/25) of current participants did not perceive relationships as imperative.

Discussion

Preliminary Findings

The primary outcome of intimate relationship dynamics (ie, imperativeness and power) is expected to be associated with high-risk sexual behaviors and having an STI, as found in populations without a history of juvenile detention in related studies. A study that included young African American women (ages 15-21), who were primarily inner-city youths seeking sexual health services from a community agency, found that participants who perceived their relationships as imperative (1) had less relationship power, (2) were more likely to perceive themselves as being unable to refuse sex, (3) were more likely to fear negotiating condom use with their partners, and (4) were more susceptible to partner abuse [21]. Furthermore, participants who perceived relationships to be imperative reported having sex while under the influence of alcohol and drugs and being willing to engage in unprotected sex [21].

Paxton et al, in a study that included African American women (ages 26-54), found that participants stayed with unfaithful partners largely due to the desire to have a relationship [24]. African American women participating in the study reported high-risk sexual behaviors, such as knowingly having sex with partners who have STIs, not requiring unfaithful partners to use condoms, only requiring unfaithful partners to use condoms with other sexual partners, and engaging in sex while high on drugs and alcohol [24]. The work of Raiford, Seth, and DiClemente [21] and Paxton et al [24] suggests that intimate relationship dynamics (imperativeness and power) are salient factors for both adolescent and adult women.

Limitations

The primary limitation is expected to be participant attrition. The population of interest for this study is housed in an RYDC, which means that it is highly fluid, resulting in recruitment challenges and a small sample size [27]. Many potential participants satisfied all inclusion criteria except for detainment length. Despite the high turnover of this population, psychosocial factors may be more salient for the RYDC group,

who have more recent free-world experiences than detainees in YDCs.

The study design only included young women currently being detained and not the perspective of young men with a history of juvenile detention. The nature of intimate relationships between young women and men, and specifically factors related to high-risk sexual behaviors, have not been well defined. This study did not seek to contact intimate partners of participants because of many potential issues, such as revisiting traumatic experiences, partners being detained/incarcerated, or relationships violating consent laws. Furthermore, few studies have investigated the dyad relationship of young women and men in regard to imperativeness and power; the majority of the studies reviewed included adolescent and adult women.

Future investigations may address high turnover and attrition by extending studies into the community for participants who are being released or who have detention durations of less than 30 days. In other words, future studies may include populations with current or past experiences with the juvenile justice system. There also may be technological tools, such as mobile applications and social media, to facilitate outreach into the community for the juvenile justice population being released. Outreach beyond detention centers also may facilitate including dyads to understand relationship dynamics more completely from the perspectives of young women and men, which may lead to more effective interventions.

Acknowledgments

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Conflicts of Interest

None declared.

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Abbreviations

DJJ: department of juvenile justice
EBI: evidence-based intervention
IPV: intimate partner violence
RYDC: regional youth detention center
STI: sexually transmitted infection
YDC: youth developmental campus

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Original Paper

Using Computational Approaches to Improve Risk-Stratified Patient Management: Rationale and Methods

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Abstract

Background: Chronic diseases affect 52% of Americans and consume 86% of health care costs. A small portion of patients consume most health care resources and costs. More intensive patient management strategies, such as case management, are usually more effective at improving health outcomes, but are also more expensive. To use limited resources efficiently, risk stratification is commonly used in managing patients with chronic diseases, such as asthma, chronic obstructive pulmonary disease, diabetes, and heart disease. Patients are stratified based on predicted risk with patients at higher risk given more intensive care. The current risk-stratified patient management approach has 3 limitations resulting in many patients not receiving the most appropriate care, unnecessarily increased costs, and suboptimal health outcomes. First, using predictive models for health outcomes and costs is currently the best method for forecasting individual patient's risk. Yet, accuracy of predictive models remains poor causing many patients to be misstratified. If an existing model were used to identify candidate patients for case management, enrollment would miss more than half of those who would benefit most, but include others unlikely to benefit, wasting limited resources. Existing models have been developed under the assumption that patient characteristics primarily influence outcomes and costs, leaving physician characteristics out of the models. In reality, both characteristics have an impact. Second, existing models usually give neither an explanation why a particular patient is predicted to be at high risk nor suggestions on interventions tailored to the patient's specific case. As a result, many high-risk patients miss some suitable interventions. Third, thresholds for risk strata are suboptimal and determined heuristically with no quality guarantee.

Objective: The purpose of this study is to improve risk-stratified patient management so that more patients will receive the most appropriate care.

Methods: This study will (1) combine patient, physician profile, and environmental variable features to improve prediction accuracy of individual patient health outcomes and costs; (2) develop the first algorithm to explain prediction results and suggest tailored interventions; (3) develop the first algorithm to compute optimal thresholds for risk strata; and (4) conduct simulations to estimate outcomes of risk-stratified patient management for various configurations. The proposed techniques will be demonstrated on a test case of asthma patients.

Results: We are currently in the process of extracting clinical and administrative data from an integrated health care system's enterprise data warehouse. We plan to complete this study in approximately 5 years.

Conclusions: Methods developed in this study will help transform risk-stratified patient management for better clinical outcomes, higher patient satisfaction and quality of life, reduced health care use, and lower costs.

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KEYWORDS

decision support techniques; patient care management; forecasting; computer simulation; machine learning

Introduction

Risk-Stratified Management of Chronic Disease Patients

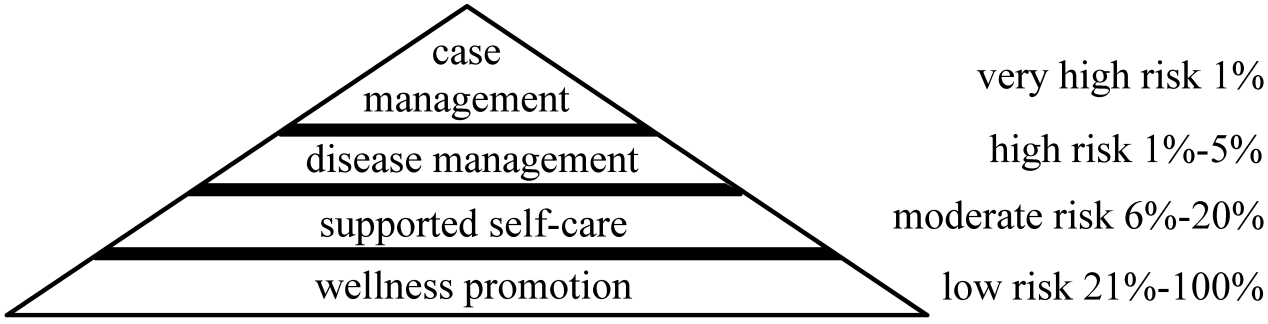
Chronic diseases affect approximately 52% of Americans and consume 86% of health care costs [1]. Example management strategies for care include case management, disease management, supported self-care, and wellness promotion (listed in Table 1 in descending order of intensity). Each strategy is widely used and has its own benefits and properties [2,3]; for example, most major employers purchase and nearly all private health plans offer case management services [2,4] targeting early interventions at high-risk patients to prevent large expenditures and avoid deterioration of health status. Proper use of case management can reduce hospital admissions and readmissions and emergency department visits by up to 30% to 40% [3,5-9], lower costs by up to 15% [6-10], and improve patient satisfaction, quality of life, and treatment adherence by 30% to 60% [5]. A case management program can cost more than US $5000 per patient per year [6] and typically enrolls only 1% to 3% of targeted patients due to resource limitations [11]. For maximal benefit, only patients expected to incur the highest costs and/or those with the poorest prognoses should be enrolled.

Table 1. Description of patient management strategies.

Management strategy	Description
Case management	"A collaborative process that assesses, plans, implements, coordinates, monitors, and evaluates the options and services required to meet [a patient's] health and human service needs" [12]. It involves a case manager who calls the patient periodically, helps make doctor appointments, and arranges for health and health-related services.
Disease management	Example intervention: check electronic medical records to find and call high-risk patients with the disease who require a specific test, but have not had it for ≥2 years.
Supported self-care	Example intervention: give patients electronic monitoring tools for self-management.
Wellness promotion	Example intervention: mail educational materials on how to maintain health.

Patients' health care use and costs have a pyramid-like distribution. A small portion of patients consume most health care resources and costs [13,14]. For instance, 25% and 80% of costs are spent on 1% and 20% of patients, respectively [11,14]. High costs often result from bad health outcomes or inappropriate use of health care. Typically, more intensive management strategies are more effective at improving health outcomes, but are also more expensive. To use limited resources efficiently, risk stratification is widely used in managing patients with chronic diseases such as asthma, chronic obstructive pulmonary disease, diabetes, and heart diseases [13]. As shown in Figure 1, available management strategies are arranged into a hierarchy [14]. Patients are stratified based on predicted risk [6] and this risk can represent either high cost or a bad health outcome. Higher risk results in more intensive care to match expected returns [15]. For example, patients with predicted risk above the 99th percentile are put into case management and so on.

Figure 1. An example hierarchy of risk-stratified management levels for chronic disease patients.



Problems With the Current Risk-Stratified Patient Management Approach

The current risk-stratified patient management approach has 3 shortcomings, which result in many patients not receiving the most appropriate care and greatly degrade its outcomes.

First, existing methods for predicting individual patients' risk have low accuracy resulting in misstratification. As shown in Allaudeen et al [16], clinicians cannot predict well which patients will become high risk in the future. Criterion-based modeling uses a priori criteria to describe high-risk patients. It is ineffective partly due to regression to the mean, in which most patients who incurred high cost or health care use in one period will stop doing so in the next period [17]. Frequently, a

predictive model for individual patient health outcome or cost is used to automatically identify high-risk patients [5,18-23]. For instance, health plans in 9 of 12 communities are reported to use predictive modeling to identify candidate patients for case management [24]. For patients with predictions of the poorest outcomes or highest costs, case managers manually review patient charts and make final management decisions. Predictive modeling greatly outperforms clinicians and criterion-based modeling [17], and is the best method for identifying high-risk patients, yet needs improvement.

Existing predictive models for individual patient health outcomes and costs have low accuracy. When predicting a patient's cost, the average error is usually as large as the average cost [25] and the R^2 accuracy measure is less than 20% [26]. When predicting a patient's health outcome, the area under the receiver operating characteristic (ROC) curve accuracy measure is often low, much less than 0.8 [27-31]. These large errors cause enrollment to align poorly with patients who would benefit most from a management program [5]. As shown in Weir et al [23], among the top 10% of patients who incurred the highest costs, more than 60% were missed in the top 10% risk group selected by a predictive model. Among the top 1% of patients who incurred the highest costs, more than 80% and approximately 50% were missed in the top 1% and 10% risk groups selected, respectively. Suppose a case management program could accommodate 1% of affected patients. Even if case managers had time to manually review the top 10% risk group selected by the model and made perfect enrollment decisions, they would still miss half of the top 1% who incurred the highest costs. The case with health outcomes is similar [29,30].

Existing predictive models primarily use patient features only, implicitly assuming that a patient's health outcome and cost depend only on the patient's characteristics and are unrelated to the treating physician's characteristics, which are influential. The use of treating physician's characteristics, or physician profile features, has been exploited minimally in predictive modeling [28] leaving a knowledge gap.

Second, patients are at high risk for different reasons. Complex predictive models, including most machine learning models such as random forest, give no explanation for a prediction of high risk. Existing models also give no suggestion on interventions tailored to the patient's specific case. An intervention addressing the reason for being at high risk tends to be more effective than nonspecific ones. For instance, for a patient who lives far from his/her physician and has difficulty accessing care, providing transportation can be effective.

A patient can be at high risk for multiple reasons each corresponding to either a single or a combination of multiple patient and physician profile features. A clinician may give the patient tailored interventions based on subjective and variable clinical judgment, but he/she is likely to miss some suitable interventions due to 3 factors:

1. Large practice variation (eg, by 1.6 to 5.6 times) exists across different clinicians, health care facilities, and regions [13,27,32-37].
2. Many features exist. A typical clinician can concurrently process no more than a single-digit number of information items [38] making it difficult to identify all these reasons due to the vast number of possible feature combinations.
3. Clinicians usually give interventions addressing patient factors only and miss those addressing physician factors. For instance, a physician may be unfamiliar with the patient's disease. Providing the physician continuing medical education on it can be effective.

Third, thresholds for risk strata are decided heuristically with no quality guarantee leading to unnecessarily increased costs and/or suboptimal health outcomes. For instance, total future cost of all patients factoring in the management programs' costs is unlikely to be minimized even under the unrealistic assumption that we know exactly (1) each patient's future risk and (2) every program's impact on each patient's future cost if the patient is put into the program. Total future cost implicitly reflects patient health outcomes and the management programs' benefits. For instance, fewer hospitalizations usually lead to lower costs.

Improving Prediction Accuracy, Explaining Prediction Results, Suggesting Tailored Interventions, and Computing Optimal Thresholds

New techniques are needed to improve risk-stratified patient management so that more patients can receive the most appropriate care. To fill the gap, we will (1) combine patient, physician profile, and environmental variable features to improve prediction accuracy of individual patient health outcomes and costs; (2) develop an algorithm to explain prediction results and suggest tailored interventions; (3) develop an algorithm to compute optimal thresholds for risk strata; and (4) conduct simulations to estimate outcomes of risk-stratified patient management for various configurations. A physician's practice profile contains his/her own information as well as clinical and administrative data of his/her patients aggregated historically. We hypothesize that using our techniques will increase prediction accuracy, improve outcomes, and reduce costs. The explanations and suggestions provided by our algorithm can help clinicians prioritize interventions and review structured attributes in patient charts more efficiently, and will be particularly useful for clinicians who are junior or unfamiliar with how to handle certain types of patients. After our methods identify patients with the highest predicted risks and give explanations and suggestions, clinicians would review patient charts, consider various factors (eg, social factors, how likely a patient's health outcome will greatly improve [39]), and make final decisions on the management levels and interventions for these patients as is often done in case management.

Innovation

This study is innovative for several reasons:

1. We will develop the first algorithm to (1) explain prediction results, which is critical for clinicians to trust the results and (2) suggest tailored interventions. Currently no algorithm can do the latter. Our algorithm will explain results for any predictive model without degrading accuracy and solve a long-standing open problem. In contrast,

existing explanation methods are usually model specific and decrease accuracy [40,41].

2. We will transform risk-stratified patient management by personalizing management strategies based on objective data. At present, clinicians give interventions based on subjective and variable clinical judgment, and miss some of the suitable interventions for many high-risk patients.
3. The added value of physician profile features in predicting health outcomes and costs has never been systematically studied. We will include physician profile characteristics to construct new features and build new predictive models accurate for individual patients.
4. To better predict individual patient costs, we will develop a new and general technique for reducing features (ie, independent variables). The technique can increase the prediction accuracy of any continuous outcome variable with a complex nonlinear relationship with many independent variables. This is particularly useful when standard feature selection techniques [42] cannot narrow down many independent variables to a few effective features.
5. We will develop the first algorithm to compute optimal thresholds for risk strata. These thresholds aim at maximizing total expected return on the entire patient population and will be better than those determined heuristically. Currently no algorithm exists for this purpose.
6. When a predictive model is used, our study will estimate outcomes of risk-stratified patient management with multiple management strategies. No such estimates have been provided before. Previous studies have estimated outcomes for a single management strategy: case management [43].
7. We will use a new simulation method to determine which attributes are the most important to include in the predictive model. Different combinations of attributes will be used to determine the minimum performance requirement and allow tradeoffs for adapting use of our models beyond our setting based on available attributes. Previous predictive models have relied on a fixed set of attributes, which may not be collected by other sites and thus do not generalize beyond the study site.
8. Often, a specific technique is useful for only a single disease or decision support application. In contrast, after proper extension, our new techniques will generalize to a variety of decision support applications and disease settings. Examples of opportunities for future studies are (1) more precise models for health outcomes and costs will augment various decision support applications for managing limited resources, such as assisting with health care resource allocation planning [44] and automatically identifying patients likely to be admitted or readmitted in the near future triggering earlier follow-up appointments or home visits by nurses to reduce admissions and readmissions; (2) adding physician profile features can improve prediction accuracy of other outcomes, such as patient satisfaction [45], patient adherence [46], and missed appointments [47], and facilitate targeting resources, such as print and telephone reminders to reduce missed appointments [47] or interventions to improve treatment adherence [46]; (3) the algorithm for explanations and suggestions can be used to explain prediction results and suggest interventions for various applications, such as to reduce missed appointments; (4) the threshold computation algorithm can help target resources for various applications; and (5) our simulation method can be used to deploy other predictive models in clinical practice.

In summary, the significance of this study is development of new techniques to help transform risk-stratified patient management and personalize management strategies so that more patients will receive the most appropriate care. Broad use of our techniques will improve clinical outcomes, patient satisfaction, and quality of life, and reduce health care use and cost.

Methods

Machine learning is a computer science area that studies computer algorithms improving automatically through experience. Machine learning methods, such as neural network, decision tree, and support vector machine, are widely used for predictive modeling [48] and will be used in our study. With less strict assumptions (eg, on data distribution), machine learning can achieve higher prediction accuracy, sometimes doubling it, than statistical methods [11,49,50].

Datasets and Test Cases

This study will use a large clinical and administrative dataset in Intermountain Healthcare's enterprise data warehouse (EDW) for all 4 aims. Intermountain Healthcare is the largest health care system in Utah, with 185 clinics and 22 hospitals. Intermountain Healthcare's EDW contains approximately 9000 tables and an extensive set of attributes [51]. Partial lists of patient and physician attributes follow.

Patient Attributes

Patient attribute data include admission date and time; age; orders (eg, medications, laboratory tests, exams, immunizations, imaging, counseling), including order name, ordering provider, performing date, and result date; allergies; barriers (eg, hearing, language, learning disability, mental status, religion, vision); cause of death; chief complaint; death date; diagnoses; discharge date; exam result; facility seen for the patient visit; gender; health insurance; health care cost (eg, billed charge, Intermountain Healthcare internal cost, and reimbursed cost); height; home address; immunizations; laboratory 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 (eg, inpatient, outpatient, urgent care, or emergency department); vital signs; and weight.

Physician Attributes

Physician attribute data include age, gender, health insurances accepted, level of affiliation with Intermountain Healthcare, office location(s), specialties, type of primary care physician, and years in practice.

Summary Statistics of the Dataset

Our contracted Intermountain Healthcare data analyst will execute Oracle database SQL queries to extract a deidentified version of the dataset, encrypt it, and transfer it securely to a password-protected and encrypted computer on which we will perform secondary analyses. Intermountain Healthcare uses dedicated tables to track changes in diagnosis and procedure codes over time. The dataset contains information on patient encounters over the past 11 years. For the last 5 years, data captured for children cover more than 400 pediatric primary care physicians, 360,698 pediatric patients (age zero to 17 years), and 1,557,713 clinical encounters per year. Data captured for adults cover more than 600 primary care physicians, 878,448 adult patients (age ≥18 years), and 5,786,414 clinical encounters per year. Asthma prevalence is approximately 7.6% in the Intermountain Healthcare pediatric population and approximately 8.6% in the Intermountain Healthcare adult population. The dataset includes approximately 400 attributes and represents electronic documentation of approximately 85% of pediatric care and approximately 60% of adult care delivered in Utah [33,52]. Intermountain Healthcare dedicates extensive resources to data accuracy and integrity. Due to its large size and attribute richness, the dataset gives us many advantages for exploring the proposed predictive models.

In addition, we will use 21 environmental variables recorded over 11 years by regional monitoring stations within the geographic area covered by Intermountain Healthcare. These variables include particulate matter up to 2.5 μm in size ($PM_{2.5}$) and 10 μm in size (PM_{10}), carbon monoxide (CO), nitrogen dioxide (NO_2), sulfur dioxide (SO_2), ozone (O_3), temperature, relative humidity, wind speed, precipitation, dew point, and activities of viruses (adenovirus; enterovirus; human metapneumovirus; influenza A virus; influenza B virus; parainfluenza virus types 1, 2, and 3; rhinovirus; and respiratory syncytial virus). Because the monitoring stations are spread across a large geographic area including the entire state of Utah, the readings of the same environmental variable can differ greatly at different monitoring stations at any time.

Using Intermountain Healthcare data, we will demonstrate our techniques on the test case of asthma patients. In the United States, asthma affects 18.7 million adults (8%) [53] and 7.1 million children (9.6%) [54,55]. Patient management strategies such as case management can ensure proper care to reduce asthma exacerbations, improve school attendance and performance, and reduce hospitalizations and emergency department visits. This impacts both quality of life and 63% of total annual asthma costs attributable to asthma exacerbations [8,56].

Our analysis results will use different combinations of attributes to determine the minimum performance requirement and allow tradeoffs for adapting use of our models beyond our setting based on available attributes. Our results will provide a cornerstone to expand testing of our techniques on other clinical datasets, patient populations, and diseases beyond asthma in the future. As patient status and feature patterns associated with high risk change over time, our techniques can be periodically reapplied (eg, to move patients across different management levels and identify newly occurring feature patterns).

Aim 1: Combine Patient, Physician Profile, and Environmental Variable Features to Improve Prediction Accuracy of Individual Patient Health Outcomes and Costs

Aim 1a: Build Predictive Models for Individual Patient Health Outcomes

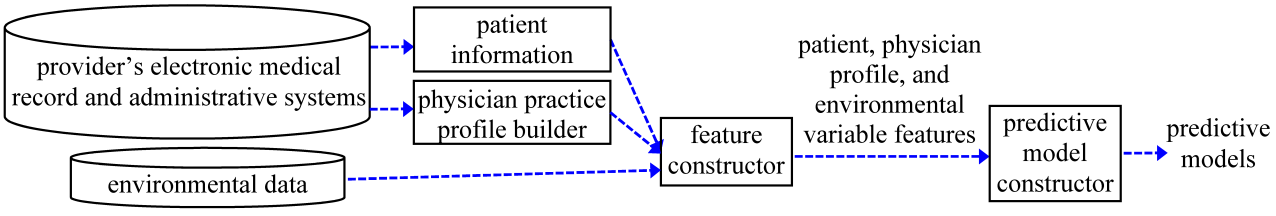
Framework

We will apply the framework shown in Figure 2 to build predictive models using patient, physician profile, and environmental variable features. Environmental variables impact outcomes of certain diseases such as asthma [57,58]. The models will be used to predict individual patient health outcomes.

For each physician, we will build a practice profile including his/her own (eg, demographic) information as well as aggregated historical information of his/her patients (excluding the index patient) from the provider's electronic medical record and administrative systems. An example physician practice profile attribute is the number of the physician's patients with a specific disease [59]. We will use patient attributes to form patient features. We will use both patient and physician practice profile attributes to form physician profile features. Each feature is formed from one or more base attributes. If the outcome variable is affected by environmental variables, we will also use environmental variable attributes to construct features. Predictive models will be built using patient, physician profile, and environmental variable features.

There are almost an infinite number of possible features. In addition, factors such as characteristics of a pediatric patient's parents can impact patient outcomes. This study's purpose is not to list all possible features, exhaust all possible factors that can affect patient outcomes, and reach the theoretical limit of maximum possible prediction accuracy. Instead, our goal is to demonstrate that adding physician profile features can improve prediction accuracy and, subsequently, risk-stratified patient management. A nontrivial improvement in health outcomes and/or reduction in costs can benefit society greatly. As is typical with predictive modeling and adequate for our targeted decision support application, our study focuses on associations.

Figure 2. A framework for building predictive models using patient, physician profile, and environmental variable features.



Data Preprocessing

We will use established techniques, such as imputation, to deal with missing values and detect and remove/correct invalid values [48,60]. For environmental variables, we will use standard methods [61,62] to obtain aggregate values, such as monthly averages, from raw values. For administrative and clinical attributes, we will use grouper models such as the Diagnostic Cost Groups system to group diseases, procedures, and drugs, and reduce features [13,25].

Patient Features

We will use standard patient features, such as age and diagnoses, that have been studied in the clinical predictive modeling literature [13,27,48]. Commonly used features are listed in Luo [32] and Schatz et al [29].

Physician Profile Features

Some physician profile features are computed using only physician practice profile attributes. Examples of such features are (1) the logarithm of the normalized number of a physician's patients with a specific characteristic, such as a specific disease, gender, race, or age range (a logarithm is used to diminish the difference in the number across physicians); (2) the logarithm of the number of specific procedures performed by a physician; (3) the mean outcome of a physician's patients with a specific disease (if a physician does not have enough patients with a specific disease, we will set the disease's mean outcome in the physician's practice profile to the mean outcome of all patients with the disease); (4) the average cost of a physician's patients with a specific disease; (5) the average ratio of chronic controller to total asthma medications of a physician's asthma patients, which is an asthma care quality measure [63-66]; (6) the mean of a feature of a physician's (pediatric) asthma patients with desirable/undesirable outcomes; (7) a physician's age; (8) a physician's total office hours per week; (9) a physician's years in practice; and (10) a physician's specialty.

Some physician profile features are formed by combining patient and physician practice profile attributes, characterizing the match of patient and physician. Examples of such features are (1) the distance between the physician's office and patient's home, (2) an indicator of whether the physician and patient are of the same gender [67], (3) an indicator of whether the physician and patient speak the same language, and (4) an indicator of whether the physician accepts the patient's insurance.

The preceding lists of physician profile features are only for illustration purposes and are by no means exhaustive. More physician profile features will be investigated in this study. When a patient is managed by multiple physicians simultaneously, the patient's outcomes are affected by the profile features of all these physicians. A traditional method for handling this situation is to use episode grouper software to split the whole span of patient care into episodes and assign each episode to a single physician [13,68]. An episode of care is "a series of temporally contiguous health care services related to treatment of a given spell of illness or provided in response to a specific request by the patient or other relevant entity" [27,69]. Apart from the episode method, we will investigate other methods to combine multiple physicians' profile features.

Environmental Variable Features

We will use standard environmental variable features such as monthly averages from clinical predictive modeling literature [57].

Definition of Asthma Cases and Outcomes

As test cases, we will focus on primary care physicians and develop and test our idea using (1) pediatric asthma and (2) adult asthma. The method described in Schatz et al [29,70,71] will be used to identify asthma patients. A patient is considered to have asthma if he/she has (1) at least one *International Classification of Diseases, Ninth Revision* (*ICD-9*) diagnosis code of asthma (493.xx) or (2) at least 2 asthma-related medication dispensing records (excluding oral steroids) in a 1-year period, including inhaled steroids, beta-agonists (excluding oral terbutaline), oral leukotriene modifiers, and other inhaled antiinflammatory drugs [29]. We will use 2 outcome measures for asthma: (1) primary outcome—whether acute care (inpatient stay, urgent care, and emergency department visit) with a primary diagnosis of asthma (*ICD-9* code: 493.xx) occurred for a patient in the following year [28,29,31,32,56,72,73] and (2) secondary outcome—the total amount of reliever medication and oral steroid medication for acute asthma exacerbations that a patient refilled in the following year. Total refill amount reflects the number and degree of asthma exacerbations experienced by the patient [63,64] and is available in our dataset.

Predictive Models

We will use Weka [74], a widely used open-source machine learning and data mining toolkit, to build predictive models. Weka integrates an extensive set of popular machine learning algorithms, ensemble techniques combining multiple predictive models, feature selection techniques, and methods for handling the imbalanced class problem. Both numerical and categorical variables appear in clinical, administrative, and environmental data. We will use supervised algorithms that can handle both types of variables, such as decision tree and *k*-Nearest Neighbor. We will test every applicable algorithm and manually tune hyperparameters.

The accuracy achieved by state-of-the-art predictive models is usually far below 80% [28,29]. We would regard Aim 1a (to build predictive models for individual patient health outcomes) partially successful if we can improve accuracy by 10% or more for either pediatric or adult asthma. We would regard Aim 1a completely successful if we can improve accuracy by 10% or more for both pediatric and adult asthma. Given a set of features, we will use 3 methods to improve model accuracy. First, some features are unimportant or highly correlated with one another, which may degrade model accuracy. To address this, we will use standard feature selection techniques, such as the information gain method, to identify important features that will be used in the model [28,42,74]. Second, for a categorical outcome variable with 2 values, the corresponding 2 classes in our dataset can be imbalanced, meaning many more instances exist for one class than the other. This can potentially degrade

model accuracy. We will use standard techniques such as Synthetic Minority Oversampling Technique (SMOTE) to address this [74]. Third, we will try ensemble techniques, such as random forest, that combine multiple models and usually work better than individual models [74].

Accuracy Evaluation and Sample Size Justification

We have 11 years' data. We will use a standard approach to train and test predictive models. We will conduct stratified 10-fold cross validation [74] on the first 10 years' data to train and estimate the accuracy of models. The 11th year's data will be used to assess the best models' performance reflecting use in practice. For categorical outcome variables, we will use the standard performance metric of the area under the curve (AUC) of the ROC [74] to select the best model. For continuous outcome variables, we will use the standard performance metric of R^2 to select the best model and also report the Cumming's prediction measure (equivalent to the mean absolute prediction error) [25,32]. To determine the clinical, administrative, and environmental variable attributes essential for high accuracy, backward elimination [48] will be used to drop independent variables as long as the accuracy does not drop by more than 0.02.

We will test the hypothesis that adding physician profile features can increase prediction accuracy twice—once for children and once for adults. We will compare the accuracies achieved by 2 predictive models using the best machine learning algorithm. The first model will use patient, physician profile, and environmental variable features; the second, only patient and environmental variable features. We will accept the hypothesis if the first model achieves higher accuracy (AUC or R^2) than the second model by 10% or more.

Consider the categorical outcome variable of acute care usage with 2 values (classes). A predictive model using only patient and environmental variable features usually achieves an AUC far less than 0.8 [28,29]. Using a 2-sided z test at a significance level of .05 and assuming for both classes a correlation coefficient of .6 between the 2 models' prediction results, a sample size of 137 instances per class has 90% power to detect a difference of 0.1 in AUC between the 2 models. The 11th year's data include approximately 27,000 children and 75,000 adults with asthma, providing adequate power for testing our hypothesis. To train a predictive model well, typically the ratio of the number of data instances to the number of features should be 10 or more. In our case, a few hundred features at most will be used; thus, our dataset would be large enough for training the predictive models. The case with the continuous outcome variable is similar (see Aim 1b: Sample Size Justification).

Aim 1b: Build Predictive Models for Individual Patient Costs.

We will use an approach similar to that in Aim 1a, but change the prediction target from health outcomes to individual patients' total costs in the following year [13,25,27]. Each medical claim is associated with a billed cost, an Intermountain Healthcare internal cost, and a reimbursed cost [13]. We will use the Intermountain Healthcare internal cost [33], which is less subject to variation due to member cost-sharing [13], and reflects actual cost more closely. To address inflation, we will standardize all costs to 2014 US dollars using the medical consumer price index [75].

In addition to the rare use of physician profile features, 2 other major reasons also cause low accuracy in predicting an individual's cost. First, most existing work on predicting costs uses linear regression models [13,25,27]. In reality, costs do not follow a linear model [26]. Second, the cost of a patient with a specific disease is the cost of treating all his/her diseases [25]. To consider this factor, each model uses many features or independent variables (eg, one feature per disease) and can easily have insufficient training data [48]. To address these 2 problems, we will try nonlinear, disease-specific, machine learning models, which were proposed in a previous paper [32], but have not been implemented so far. This method's key idea is to reduce features by merging several less important features into one feature while maintaining important features as separate. The current approach of identifying important features and grouping other features is manual. We will also investigate automatic approaches. For example, we can regard the top features with the largest associations with the outcome variable as important ones. The remaining features are clustered using a similarity metric to form groups. The automatic approach is general and can be used to improve prediction accuracy of any continuous outcome variable that has a complex nonlinear relationship with many independent variables.

Sample Size Justification

In predicting an individual's cost, a predictive model using only patient and environmental variable features usually achieves an R^2 <20% [26]. Using an F test at a significance level of .05 and assuming the presence of 70 patient and environmental variable features, a sample size of 245 patients has 90% power to detect an increase of 10% in R^2 attributed to 30 physician profile features. The 11th year's data include approximately 27,000 children and 75,000 adults with asthma, providing adequate power for testing our hypothesis of an increase of 10% or more in R^2.

Our goal is to achieve a 10% or more improvement in accuracy. If our models cannot achieve high accuracy on the entire group of asthma patients, we will build separate models for different subgroups of asthma patients. Patient subgroups are defined by specific characteristics, such as age, prematurity, comorbidity, or insurance type that are usually independent variables of the original models. If our models still cannot achieve high accuracy, we will conduct subanalyses to identify patient subgroups on which our models perform well. In this case, our final models will be applied only to the identified patient subgroups.

A missing data problem occurs when a patient has several physicians belonging to different provider groups, with no single provider having complete information on the patient. We anticipate that adding physician profile features can improve prediction accuracy even if some data are missing. The missing data problem is unlikely to be an issue for children in our case, as Intermountain Healthcare provides approximately 85% of pediatric care in Utah [52]. If the Intermountain Healthcare

EDW misses too much data for adults, we will use claim data in the all-payer claims database [76] to compensate. In the future when applying our predictive models to other health care systems, this compensation strategy can be used. Also, we expect missing data problems to be uncommon in health maintenance organization settings where all physicians managing the patient belong to the same provider group and the provider's electronic medical record and administrative systems usually have all medical data collected on the patient [77].

As mentioned previously, identifying asthma requires medication order and refill information. Our dataset includes this information because Intermountain Healthcare has its own health insurance plan (SelectHealth [78]). If the Intermountain Healthcare EDW is missing too much refill information, we will use claim data in the all-payer claims database [76] to compensate. If adding physician profile features cannot significantly increase prediction accuracy for asthma, we will choose chronic obstructive pulmonary disease or heart diseases for Aims 1 to 4.

We have a large dataset. If we experience scalability issues using Weka, we will use a parallel machine learning toolkit, such as Spark's MLlib [79-81], to build predictive models on a secure computer cluster available to us at the University of Utah Center for High Performance Computing [82].

Aim 2: Develop an Algorithm to Explain Prediction Results and Suggest Tailored Interventions

For patients with predicted risk greater than a predetermined threshold, such as the 95th percentile, this aim will explain prediction results and suggest tailored interventions. These explanations and suggestions can help clinicians make final decisions on the management levels and interventions for these patients.

Prediction accuracy and model interpretability are frequently 2 conflicting goals. A model achieving high accuracy is usually complex and difficult to interpret. How to achieve both goals simultaneously has been a long-standing open problem. Our key idea to solve this problem is to separate prediction and explanation by using 2 models concurrently, each for a different purpose. The first model makes predictions and targets maximizing accuracy. In this study, this model is the best one built for the outcome variable in Aim 1. The second model is rule-based and easy to interpret. It is used to explain the first model's results rather than make predictions. The rules used in the second model are mined directly from historical data rather than coming from the first model. For each patient whom the first model predicts to be at high risk, the second model will show zero or more rules. Each rule gives a reason why the patient is predicted to be at high risk. Because some patients can be at high risk for rare reasons that are difficult to identify, we make no attempt to ensure that at least one rule will be shown for every patient predicted to be at high risk. Instead, we focus on common reasons that are more important and relevant to the patient population than rare ones. We expect most high-risk patients to be covered by one or more common reasons.

We will use an associative classifier [83-85] from the data mining field as the second model. Associative classifiers can handle both numerical and categorical variables and be built efficiently from historical data. Compared with several other rule-based models, an associative classifier includes a more complete set of interesting and useful rules and can better explain prediction results. For ease of description, our presentation focuses on the case that each patient has exactly one data instance (row). The case in which a patient has more than one data instance can be handled similarly. We will proceed in 3 steps.

In step 1, association rules are mined from historical data. As mentioned in Aim 1, each patient is described by the same set of patient, physician profile, and environmental variable features and labeled as either high risk or not. An associative classifier includes a set of class-based association rules. Each rule includes a feature pattern associated with high risk and is of the form: p_1 AND p_2 AND ... AND p_k is associated with high risk. The value of k varies across different rules. Each item p_i ($1 \le i \le k$) is a feature-value pair of the form (f, v) indicating that feature f takes a value equal to v (if v is a value) or within v (if v is a range). The rule suggests that a patient is likely to be at high risk if he/she satisfies p_1, p_2, ... , and p_k. An example rule is the patient was hospitalized for asthma last year AND the patient's primary care physician has fewer than 10 asthma patients is associated with high risk.

For a given association rule, the percentage of patients satisfying the rule's left side and being at high risk reflects the rule's coverage and is called the rule's *support*. Among all patients satisfying the rule's left side, the percentage of patients at high risk reflects the rule's accuracy and is called the rule's *confidence*. An associative classifier includes association rules at a given level of minimum support (eg, 1%) and confidence (eg, 70%). These rules can be efficiently mined from historical data using existing techniques [83-85], which can eliminate redundant and noisy rules. Because we need only rules suggesting high risk, we can mine desired feature patterns (ie, the rules' left side) from high-risk patients' data rather than from all patients' data to improve the efficiency of rule generation.

Typically, many association rules will be mined from historical data [83-86]. Keeping all these rules will overwhelm clinicians. To address this issue, we will use 3 methods to reduce the number of rules. First, in forming rules, we will consider only features appearing in the first model that is used to make predictions. As mentioned in Aim 1a, many nonessential features will be removed during feature selection and backward elimination when building the first model. Second, we will focus on rules with no more than a predetermined small number of items (eg, 4) because long feature patterns are difficult to understand and act on [83]. Third, users can optionally specify for a feature what values or type of range (eg, stating that the feature is above a threshold) may potentially indicate high risk and appear in rules [40,87]. The other values or types of range are not allowed to appear in rules. This also helps form clinically meaningful rules.

In step 2, interventions will be listed for the mined association rules. Through discussion and consensus, our clinical team will examine mined association rules and remove those that make little or no clinical sense. For each remaining rule, the clinicians will list zero or more interventions addressing the reason given by the rule. Example interventions for patients include (1) provide transportation or telemedicine for a patient living far from his/her physician, (2) schedule longer or more frequent doctor appointments for a patient with multiple comorbidities, (3) schedule appointments with nurse educators or clinical pharmacists for a patient with multiple comorbidities, (4) arrange language service for a doctor appointment if the patient and physician speak different languages, and (5) give wearable air purifiers to certain types of asthma patients living in an area with bad air quality.

Example interventions at the system level include (1) provide the primary care physician continuing medical education on a specific disease, cultural competence, women's health, or pediatric health if he/she is unfamiliar with or cannot well manage the disease, patients of a particular race, diseases in women, or pediatric diseases (a physician may be unfamiliar with a disease if he/she has few patients with it; a bad mean outcome of a physician's patients with the disease may indicate, but not always, that the physician cannot manage the disease well); (2) extend physician office hours; and (3) open a new primary care clinic in an area with no such clinic nearby.

Interventions for patients are displayed to clinicians in step 3. Interventions at the system level are optional and may be viewed only by managers of the health care system. We call a rule actionable or nonactionable based on whether or not at least one intervention is associated with it. The remaining rules and their associated interventions will be stored in a database to facilitate reuse.

In step 3, prediction results are explained and tailored interventions are suggested. At prediction time, for each patient identified as high risk by the first model, we will find all association rules whose left side is satisfied by the patient using an index for rules [84]. We will display the actionable rules above the nonactionable ones, each in descending order of confidence [84]. If 2 rules have equal confidence, the rule with higher support will be ranked higher. If 2 rules have the same confidence and support, the one with fewer items will be ranked higher. Our rule sorting method differs from several traditional ones [83-85] because our goal is to explain the prediction result for a patient rather than to maximize the average prediction accuracy in a patient group. We will list confidence and associated interventions, if any, next to each rule to help the clinician identify suitable tailored interventions. By default, we will show no more than a predetermined small number of rules (eg, 3). If desired, the clinician can opt to view all rules applicable to the patient.

Commonly used support and confidence thresholds [83-85] may not be suitable for our case, in which only a small percentage of patients are at high risk. We will adjust the support and confidence thresholds if the commonly used ones cannot produce enough meaningful association rules. By setting the thresholds low enough, we will produce meaningful rules at the expense of our clinicians spending time removing rules that make little or no clinical sense. Because existing predictive models give no suggestion on tailored interventions, we will regard Aim 2 successful if a nontrivial percentage (eg, ≥20%) of high-risk patients are covered by actionable rules.

Performance Evaluation

The algorithm for explanations and suggestions will be evaluated in Aim 4.

Aim 3: Develop an Algorithm to Compute Optimal Thresholds for Risk Strata

In risk-stratified management, chronic disease patients are stratified into multiple levels [14,15]. This aim will compute the optimal thresholds for these levels that minimize total future cost of all patients factoring in the management programs' costs. Total future cost implicitly reflects patient health outcomes, health care use, efficiency of care, and the management programs' benefits. For instance, fewer hospitalizations usually lead to lower costs. The following discussion focuses on stratification based on predicted patient risk of experiencing a specific type of undesirable event (eg, hospitalization or emergency department visit). The case of stratification based on predicted cost or with more than one type of undesirable event can be handled similarly. Our discussion applies to any predictive model and is based on a fixed period in the future, such as the next 12 months.

Threshold Computation Algorithm

We will conduct quantitative analysis to determine the optimal management level for each risk percentile. We will proceed through the risk percentiles one by one, from the highest to the lowest. Given a risk percentile, we will compute for each management level the average future cost per patient in the percentile if patients in the percentile are put into the level. The level with capacity remaining in its management program and the lowest average future cost per patient will be chosen for the risk percentile.

More specifically, consider a risk percentile and an average patient whose predicted risk falls into the percentile. If the patient is enrolled in a management program, we estimate that the patient's future cost will change by delta = the program's cost − the program's benefit gained by reducing undesirable events=c_i–avg_n_e*p*c_e compared with no enrollment. Here, c_i is the program's average cost per patient. Factors such as increased medication cost due to better medication adherence are included in c_i. avg_n_e is the average number of undesirable events that a patient in the risk percentile will experience in the future. p is the percentage of undesirable events the management program can help avoid, reflecting the program's benefit. c_e is the average cost of experiencing the undesirable event once. c_i and p can be obtained from statistics reported in the literature for the management program [39,88]. avg_n_e can be obtained by making predictions on historical data and checking the corresponding statistics for the risk percentile. c_e is obtained from statistics on historical data. The management level with the smallest delta is optimal for the risk percentile. If no statistics on c_i and p of a management program are available in the

literature, the clinician in our research team (Dr Stone) will provide rough estimates based on experience. We will perform sensitivity analysis when choosing thresholds by varying the estimated values of c_i and p to obtain the full spectrum of possible outcomes in Aim 4.

The preceding method performs an exhaustive search among all management levels for each risk percentile. In practice, we would expect avg_n_e to decrease as the predicted patient risk of experiencing undesirable events becomes smaller. We will investigate using this property to reduce the search space when going through the risk percentiles one by one, from the highest to the lowest.

Performance Evaluation

The threshold computation algorithm will be evaluated in Aim 4.

Aim 4: Conduct Simulations to Estimate Outcomes of Risk-Stratified Patient Management for Various Configurations

To determine a predictive model's value for future deployment in clinical practice, we need to estimate outcomes of risk-stratified patient management when the model is used and determine how to generalize the model to differing sites collecting different sets of attributes. Our models will be built on Intermountain Healthcare datasets. Our simulations will guide how to deploy the models in another health care system. No previous study has either estimated outcomes for a model with more than one management strategy or determined the attributes most important for generalizing the model. We will demonstrate our simulation method for the task of risk-stratified management of (1) asthmatic children and (2) asthmatic adults by using our models for predicting acute care use for asthma in the following year (see Aim 1a: Definition of Asthma Cases and Outcomes), the hierarchy of risk-stratified management levels shown in Figure 1, and our algorithms described in Aims 2 and 3. Our simulation method is general and can be used to deploy other models in clinical practice. We will first evaluate the technique in Aim 1.

Outcomes

We will focus on the outcomes of costs, hospital admissions, and emergency department visits in the following year. Cost is the primary outcome, reflecting health care use and efficiency of care. Other outcomes are secondary and are indirectly reflected in costs.

Estimate Outcomes

Given a set of attributes and a predictive model, we will estimate each outcome. We will use the same method as in Aim 1 to train the model on the first 10 years' data. For the 11th year's data, we will obtain prediction results, compute thresholds for risk strata, then estimate the outcome in a way similar to Aim 3. For example, consider a patient who will have a cost of h and experience n_e undesirable events in the following year with no program enrollment. If the patient is enrolled in a management program, we estimate that the patient's future cost will become $h + c_i – n_e*p*c_e$, where c_i, p, and c_e are as defined in Aim 3. The overall outcome estimate is the aggregate of estimated outcomes for all patients. Using a similar approach, we can identify the minimum accuracy requirement of the model for it to be clinically valuable.

Sensitivity Analysis

Intermountain Healthcare collects an extensive set of attributes. Another health care system may collect only a subset of these attributes. To ensure the model's generalizability, we will test various combinations of attributes and estimate outcomes when the modified model is used. The estimate will identify which attributes are critical. If an important attribute is unavailable in a specific health care system, the estimate can suggest alternative attributes with minimal negative impact on outcomes.

Our full model will use up to 400 attributes. It is not possible to conduct simulations for every possible combination of these attributes. Instead, we will use an attribute grouping approach associating attributes likely to coexist, such as attributes associated in a laboratory test panel, based on our clinical expert's judgment. We will construct and publish a table listing possible combinations of attributes by groups, including outcomes estimated through simulations and the predictive model's trained parameters. A health care system interested in deploying the model can use the table to determine expected outcomes for their data environment and identify attributes that need to be collected. One entry in the table will correspond to the attributes available in the Observational Medical Outcomes Partnership (OMOP) common data model [89], which standardizes clinical and administrative attributes from more than 10 large health care systems in the United States [90]. The model in this entry will directly apply to at least those health care systems. If conducting simulations for the many combinations of attribute groups is too slow on one computer, we will parallelize simulations on a secure computer cluster available to us [82].

Outcome Evaluation and Sample Size Justification

We will compare outcomes achieved by 2 predictive models using the best machine learning algorithm. The first model will use patient, physician profile, and environmental variable features; the second only patient and environmental variable features. We will test 3 hypotheses: adding physician profile features will be associated with reduced (1) costs, (2) hospital admissions, and (3) emergency department visits. We will test each hypothesis twice, once for children and once for adults. Cost data will be log-transformed due to skewed distribution [13]. We will accept the primary hypothesis if the first model can reduce the log cost by 10% multiplied by its standard deviation compared with the second model. One-sided paired-sample t test will be used to test the difference in log cost between the 2 models' outcomes. McNemar's test will be used to test the difference in hospital admissions and emergency department visits. At a significance level of .05, a sample size of 857 instances has 90% power to confirm the primary hypothesis. The 11th year's data include approximately 27,000 children and 75,000 adults with asthma, providing adequate power for testing the primary hypothesis.

We will do 2 similar analyses to compare our threshold computation algorithm versus the current method of determining thresholds heuristically (evaluate the technique in Aim 3) and our algorithm for explanations and suggestions versus the current method of giving no explanation and suggestion (evaluate the technique in Aim 2). Physician profile features will be used in either analysis. In the first analysis, we will use the heuristically determined thresholds reported in the literature [15]. In the second analysis, we will use our threshold computation algorithm and estimate outcomes of our algorithm for explanations and suggestions. For an intervention, we will use statistics on its benefits and average cost per patient from the literature [39] where available. If no information is available, the clinician in our research team (Dr Stone) will conservatively estimate these numbers' minimum and maximum values based on experience. For each number, we will use 5 levels ranging from the minimum to the maximum value. To obtain the entire spectrum of possible outcomes, we will perform sensitivity analysis by varying the level and percentage of suggested interventions that clinicians will use. For the current method of giving no explanation and suggestion, we will proceed in a similar way by letting Dr Stone estimate the lower and upper bounds of the likelihood that clinicians will use an intervention. If Dr Stone has difficulty estimating the likelihood that clinicians will use an intervention, we will interview clinicians using sample patient cases to help with the estimation. Based on its own estimate of the situation, a health care system can check where in the spectrum it will fall.

Ethics Approval

We have already obtained institutional review board approvals from the University of Utah and Intermountain Healthcare for this study.

Results

We are currently in the process of extracting clinical and administrative data from the Intermountain Healthcare EDW. We plan to complete this study in approximately 5 years.

Discussion

Our techniques' principles are general and rely on no special property of any disease, patient population, or health care system. Just as predictive models are used for case management for various diseases and patient populations [13,24,30,31], after proper extension our techniques can be used for a range of decision support applications in various settings (see the innovation subsection of the Introduction). Our simulation method will determine how to generalize a predictive model to differing sites collecting different sets of attributes and the attributes most important for generalization. Using data from an integrated health care system with many heterogeneous facilities spread over a large geographic area, we will demonstrate our techniques on the test case of asthma patients. These facilities include 22 hospitals and 185 clinics, ranging from tertiary care hospitals in metropolitan areas staffed by subspecialists to community urban and rural clinics staffed by family physicians and general practitioners with limited resources. Variation in geographic location, patient population, cultural background, staff composition, and scope of services provides a realistic situation to identify factors generalizable to other facilities nationwide. When conducting simulations for each disease (pediatric/adult asthma), one of the models produced will directly apply to 10 or more large health care systems.

Because inaccurate predictive models are commonly used already for case management [24], we would expect our more precise models to have practical value. Future studies will demonstrate our techniques on other diseases, test cases, and patient populations, implement our techniques in a major health care system for risk-stratified management of asthmatic children, and test the impact in a randomized controlled trial.

In summary, our work will transform risk-stratified patient management and personalize management strategies based on objective data so that more patients will receive the most appropriate care. This will improve clinical outcomes and reduce health care use and cost. We will achieve generalizable advances in predictive modeling, explaining prediction results, tailoring interventions, and resource allocation. After proper extension, our new techniques can be used for a variety of decision support applications in various disease settings. The new simulation method will be useful for estimating outcomes for a predictive model in dissimilar data environments.

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Authors' Contributions

GL had primary responsibility for the manuscript. He conceptualized and designed the study, conducted literature review, and drafted the manuscript. BS, FS, and MM contributed to conceptualizing the presentation, gave feedback on miscellaneous medical issues, and revised the manuscript. XS participated in conceptualizing and writing the statistical analysis sections. All authors read and approved the final manuscript.

Conflicts of Interest

None declared.

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Abbreviations

AUC: area under the curve
EDW: enterprise data warehouse
ICD-9: International Classification of Diseases, Ninth Revision
OMOP: Observational Medical Outcomes Partnership
ROC: receiver operating characteristic
SMOTE: Synthetic Minority Oversampling Technique

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Original Paper

ProKaSaRe Study Protocol: A Prospective Multicenter Study of Pulmonary Rehabilitation of Patients With Sarcoidosis

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Abstract

Background: Available data assessing the efficacy of pulmonary rehabilitation for patients with chronic sarcoidosis are scant; for Germany, there are none at all.

Objective: To gain information about the benefit of in-house pulmonary rehabilitation for patients with chronic sarcoidosis and for the health care system, we intend to collect data in a prospective multicenter “real-life” cohort trial.

Methods: ProKaSaRe (Prospektive Katamnesestudie Sarkoidose in der pneumologischen Rehabilitation) [Prospective Catamnesis Study of Sarcoidosis in Pulmonary Rehabilitation] will assess a multimodal 3-week inpatient pulmonary rehabilitation program for adult patients with chronic sarcoidosis over a 1-year follow-up time. Defined specific clinical measurements and tests will be performed at the beginning and the end of the rehabilitation. In addition, questionnaires concerning health-related quality of life and the patients’ symptoms will be provided to all patients. Inclusion criteria will be referral to one of the 6 participating pulmonary rehabilitation clinics in Germany for sarcoidosis and age between 18 and 80 years. Patients will only be excluded for a lack of German language skills or the inability to understand and complete the study questionnaires. To rule out seasonal influences, the recruitment will take place over a period of 1 year. In total, at least 121 patients are planned to be included. A descriptive statistical analysis of the data will be performed, including multivariate analyses. The primary outcomes are specific health-related quality of life (St George’s Respiratory Questionnaire) and exercise capacity (6-minute walk test). The secondary outcomes are several routine lung function and laboratory parameters, dyspnea scores and blood gas analysis at rest and during exercise, changes in fatigue, psychological burden, and generic health-related quality of life (36-item Short Form Health Survey).

Results: Funding was obtained on October 12, 2010; enrollment began on January 15, 2011 and was completed by January 14, 2012. Results are anticipated late summer 2015.

Conclusions: Due to the large number of participants, we expect to obtain representative findings concerning the effectiveness of pulmonary rehabilitation for patients with sarcoidosis and to provide a dataset of assessed objective and subjective short- and long-term changes due to pulmonary rehabilitation. The results should form the basis for the planning of a randomized controlled trial.

Trial Registration: German Clinical Trials Register: DRKS00000560; https://drks-neu.uniklinik-freiburg.de/drks_web/navigate.do?navigationId=trial.HTML&TRIAL_ID=DRKS00000560 (Archived by WebCite® at http://www.webcitation.org/6dKb5X87R)

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KEYWORDS

sarcoidosis; rehabilitation; quality of life; fatigue

Introduction

Sarcoidosis is a granulomatous systemic disease of still unknown etiology [1] that seemingly is caused by a combination of genetic and environmental factors [2]. The lungs and thoracic lymph nodes are impaired in approximately 90% of patients [3,4]. Because of the systemic nature of the disease, any organ (eg, eyes, heart, skin, kidneys, liver, and the central nervous system) can be affected [5]. Due to its various manifestations, sarcoidosis shows very heterogeneous clinical pictures [6-8].

The incidence and prevalence of sarcoidosis have not been precisely compiled for Germany. Estimates using data from the Bonn Sarcoidosis collective give a prevalence of approximately 44 per 100,000 inhabitants [9]. Sarcoidosis may occur at any age, although the age distribution peaks between 20 and 40 years; women are more affected than men [9]. Therapeutic strategies aim to reduce the progressing sarcoidosis symptoms. Spontaneous remissions occur in a high percentage of cases. Nevertheless, more than 20% of sarcoidosis patients will develop a chronic or recurrent form of the disease; up to 10% suffer serious complications such as lung fibrosis or cardiac symptoms. Unfortunately, up to 5% of all cases end fatally [9].

Depending on the affected organs, sarcoidosis may cause functional limitations and reduce quality of life. It is mostly accompanied by a pronounced fatigue [9-11] and myasthenia [12]. Systemic corticosteroids remain the treatment of choice when organ function is threatened or progressively impaired [13,14]. The muscle atrophy in sarcoidosis patients is the main reason why, in addition to medication, a rehabilitation program with particular emphasis on training [15] in a multidisciplinary treatment and management setting is strongly recommended for sarcoidosis patients [11,16-19]. In contrast to rehabilitation for patients with chronic obstructive pulmonary disease (COPD), the available data assessing the efficacy of pulmonary rehabilitation for patients with sarcoidosis are sparse; for Germany, there are no current data available at all [19-21]. Therefore, we intend to close this gap by collecting data in a prospective multicenter study, which should act as a pilot for the planning of a randomized controlled trial (RCT).

Rehabilitation in Germany is a statutorily regulated part of the health care system and is only approved when social participation and inclusion, daily life, and the professional activities of the patient are restricted by illness. Access to inpatient rehabilitation is only possible following a thorough investigation by physicians specializing in social medicine. Proof that all outpatient treatment options have been exhausted must also be provided.

ProKaSaRe (Prospektive Katamnesestudie Sarkoidose in der pneumologischen Rehabilitation) [Prospective Catamnesis Study of Sarcoidosis in Pulmonary Rehabilitation] is a “real-life” cohort trial and a prospective multicentric follow-up (catamnesis) study of sarcoidosis in pneumological rehabilitation. It is designed to assess the impact of a multimodal 3-week inpatient pulmonary rehabilitation on symptoms, exercise capacity, and health-related quality of life (HRQL) for adult patients with chronic sarcoidosis. ProKaSaRe has a 1-year follow-up time.

We intend to rule out seasonal influences by including patients over a recruitment phase of 12 months. By assessing changes over a long period in this cohort of chronic sarcoidosis patients, so far the largest worldwide, we hope to gain reliable insights into and sound evidence of objective and/or subjective short- and long-term health changes caused by pulmonary rehabilitation. The most important questions that should be answered with the ProKaSaRe study are:

1. What are the characteristics of patients treated in inpatient pulmonary rehabilitation?
2. What are the short- and the long-term findings in view of symptoms, exercise capacity, and HRQL?
3. Will the HRQL change after the patients are discharged and how will the HRQL develop in the 12 months following rehabilitation?

The aim of the ProKaSaRe study is to create a solid basis of empirical real-life data on the effectiveness of pulmonary rehabilitation for patients with chronic sarcoidosis, including clinical parameters, exercise capacity, quality of life, and psychological burden via a prospective, multicentric, real-life observational study.

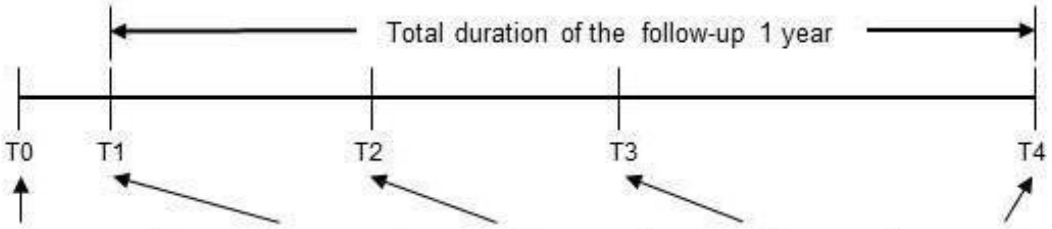
Methods

Design

ProKaSaRe is a prospective, multicentric observational study. All participants will be observed undergoing the usual structured, multidisciplinary rehabilitation program (Figure 1), which routinely includes a diagnostic assessment consisting of lung function measurements, a 6-minute walk test (6MWT) and various laboratory tests (described subsequently). As part of the study, patients with chronic sarcoidosis will be asked to answer several questionnaires at the beginning and at the end of the usual pulmonary rehabilitation program, as well as after 3, 6, and 12 months.

Ethical approval was granted by the Ethical Committee of the Hannover Medical School, Hannover, Germany (Nr 877).

Figure 1. Checkpoints for the different tests, measurements, and distribution of the questionnaire.



T_0 Start of the rehabilitation program	T_1 End of the rehabilitation program	T_2 3 months after rehabilitation program	T_3 6 months after rehabilitation program	T_4 12 months after rehabilitation program
1. Core data				
2. Diagnoses and proof				
3. Organ involvement and general symptoms				
4. Lung function	4. Lung function			
5. Gas exchange	5. Gas exchange			
6. Physical fitness	6. Physical fitness			
8. Laboratory parameters	8. Laboratory parameters			
11. Sociomedical status	11. Sociomedical status			
	13. Evaluation of rehabilitation program			
	14. Components of the individual rehabilitation program			
		12. Sociomedical patient questionnaire	12. Sociomedical patient questionnaire	12. Sociomedical patient questionnaire
7. Modified Medical Research Council Dyspnea Scale	7. Modified Medical Research Council Dyspnea Scale	7. Modified Medical Research Council Dyspnea Scale	7. Modified Medical Research Council Dyspnea Scale	7. Modified Medical Research Council Dyspnea Scale
9. Health related quality of life	9. Health related quality of life	9. Health related quality of life	9. Health related quality of life	9. Health related quality of life
10. Psychological distress	10. Psychological distress	10. Psychological distress	10. Psychological distress	10. Psychological distress
15. Response shift	15. Response shift	15. Response shift	15. Response shift	15. Response shift

Selection of Study Centers and Participants

Rehabilitation clinics specializing in pulmonary rehabilitation of sarcoidosis patients that have already showed interest during a planning conference initiated by the Bad Reichenhall clinic, Bad Reichenhall, Germany, will be formally invited to participate in the trial. All centers have proven sarcoidosis expertise and the selection will be made so that a variety of differing organizational structures, including private clinics and clinics under the auspices of the German statutory pension insurance scheme will be represented.

Based on the referral rate of previous years, we estimate that the participation of 6 pulmonary rehabilitation–specialized centers will be necessary to recruit a sufficient number of patients in 12 months to perform meaningful statistical data analysis.

To be eligible for inclusion in the ProKaSaRe study, patients must be referred for sarcoidosis to one of the 6 participating study centers for inpatient pulmonary rehabilitation by their physicians or by the hospital doctors following acute inpatient hospital treatment. If the leading rehabilitation diagnosis of sarcoidosis is confirmed by a pneumologist expert in sarcoidosis

on admission to the rehabilitation program, every patient who is younger than 80 years and aged at least 18 years will be invited to participate in the trial. On receipt of written consent, the patients will be consecutively recruited to and enrolled in the study regardless of their type of health insurance to assess a representative study population.

Patients will only be excluded for poor German language skills, the inability to understand and complete the study questionnaires (due to lack of linguistic proficiency), or if no written consent can be obtained.

Data Collection and Compilation

Data will be collected at 5 time points: T_0-T_4. Defined specific medical measurements and tests will be undertaken at the beginning and the end of the pulmonary rehabilitation program and a questionnaire concerning symptoms and HRQL will be provided at different points in time during the inpatient program and the follow-up period of 12 months (Table 1). Clinical data, sociodemographic data, medication and comorbidities, generic and illness-specific quality of life questionnaires and validated questionnaires documenting anxiety and depression, fatigue, and dyspnea will be monitored on admission at the start (T_0) and at discharge from the rehabilitation program (T_1) [22].

At T_0, the core data will be compiled comprising sociomedical status, age, sex, body mass index (BMI), severity of sarcoidosis, and organ involvement and damage. Previous treatment will also be assessed.

The following clinical findings will be recorded by the doctors at the start (T_0) and end (T_1) of the rehabilitation program at each clinic using medical survey forms: lung function, gas exchange, physical fitness, and laboratory parameters. This includes the following measurements:

1. BMI [23]
2. Lung function values: vital capacity (VC), residual volume (RV), total lung capacity (TLC), forced expiratory volume in 1 second (FEV_1), specific airway resistance (sRaw), mouth occlusion pressure 0.1 seconds after onset of inspiration (P0.1), and maximal inspiratory mouth pressure (Pimax), and Tiffeneau-Index (the FEV_1/VC ratio) [24]
3. Gas exchange: blood gas analysis at rest and during exercise (25/50 Watt), transfer factor of the lung for carbon monoxide (Tlco) in a single breath (Tlco SB) and per alveolar volume (Tlco/Va)
4. Physical fitness: 6MWT distance [25]
5. Laboratory parameters: angiotensin-converting enzyme (ACE) and C-reactive protein (CRP)

The psychological burden will be also documented at T_0 using the Hospital Anxiety and Depression Scale (HADS-D) [26] and quality of life by the 36-item Short Form Health Survey (SF-36) and the St George's Respiratory Questionnaire (SGRQ) [27]. The fatigue symptoms, which are often predominant in sarcoidosis, will be assessed by the Fatigue Assessment Scale (FAS) [28,29] and dyspnea will be measured using the Modified Medical Research Council Dyspnea Scale (mMRC)[30-32]. At 3, 6, and 12 months (T2, T3, and T4) after the rehabilitation program, the FAS, HADS-D, mMRC [30], SF-36, and SGRQ questionnaires will be completed again by the patients. Although the questionnaires at T_0 and T_1 will be submitted on site, the ones at T_2 to T_4 will be sent by post. The patients will be instructed to complete all questionnaires independently.

In addition, the patients' views of the components of the individual rehabilitation programs and their perceived importance will be assessed at discharge (T_1). Moreover, an evaluation of the program from the patients' perspective will be included in the final questionnaire at T_4.

During the whole study, response shift will be assessed by a questionnaire comprising 5 questions illustrating the prospective hopes of the patient and the retrospective specific burden of disease on the patient.

Data collection by post at 3, 6, and 12 months after discharge from the clinics will proceed by means of standardized letters with enclosed questionnaires and stamped return envelopes. The completed questionnaires will be returned to the clinic at which the rehabilitation program was carried out. Nonresponding patients will be reminded to answer the questionnaire by an additional letter after 2 weeks. Further details and the distribution times of the questionnaires and the documentation of their components are shown in Figure 1 and Table 1.

Table 1. Components of the questionnaires given at the beginning of rehabilitation (T_0), at the end of rehabilitation (T_1), and at 3, 6, and 12 months after the end of rehabilitation (T_2, T_3, and T_4).

Component	Content
1. Core data	Age, sex
2. Diagnoses	How they were secured or their positive proof
3. Organ involvement and general symptoms	
4. Lung function tests	VC, RV, TLC, FEV_1, sRaw, Tiffeneau-Index (each before/after bronchospasmolysis), P0.1, and Pimax (T_0, T_1)
5. Gas exchange	Resting and exercise blood gas analysis (25/50 Watt), Tlco SB, Tlco/VA, (T_0, T_1)
6. 6MWT	6-minute walk test distance
7. mMRC	The Modified Medical Research Council Dyspnea Scale
8. Laboratory parameters	ACE, CRP
9. Health-related quality of life	SGRQ and SF-36
10. Psychological burden	HADS and FAS
11. Sociomedical status	Standardized questionnaire (patients' self-development)
12. Sociomedical patient questionnaire	Standardized questionnaire (patients' self-development)
13. Evaluation of rehabilitation program	Evaluation of the rehabilitation program from the perspective of the patient
14. Components of rehabilitation program	Components of the individual rehabilitation program and its (patient) attributed value
15. Response shift	Prospective hopes and the retrospective specific burden of disease

All questionnaires and documents will be anonymized at the 6 participating rehabilitation centers and sent to the Hannover Medical School in Hannover, Germany, for digitalization via independent double data entry by 2 individuals who are not involved in patient care.

Sample Size Calculation and Statistics

Sample size calculation was carried out separately for both primary endpoints. Because both endpoints are equally important, the type I error was adjusted by Bonferroni multiplicity adjustment.

The minimal clinical difference for the health-related SGRQ corresponds to a change of 4 points in the score for patients with COPD. This value was also used in the planning of this study because no sarcoidosis-specific reference values were available. According to Jones [33], a 4-point difference between baseline values and follow-up data would indicate clinically relevant (long-term) effects. From the 95% confidence interval he observed in 97 patients, we estimated a standard deviation of 12.1. Assuming a 2-sided type I error of 2.5% and a power of 90%, the overall sample size required based on a paired *t* test would be 115 patients.

One estimated minimal clinical difference for the 6MWT distance corresponds to 26 m [34]. In absence of sarcoidosis-specific references, we suggest according to Marcon [35], a 6MWT distance of approximately 600 m (SD 80). Assuming a 2-sided type I error of 2.5% and a power of 90%, the overall sample size on the basis of a paired *t* test is 121. Thus, at least 121 patients are needed to test for both primary endpoints. To avoid a systematic "center bias," each center should recruit at least 30 patients. Sample size calculations were performed using nQuery Advisor7.0.

Outcome Measurement

As the primary outcome, the following parameters are defined: (1) HRQL determined using the SGRQ [25,33,36] at T_1 to T_4 and (2) exercise capacity / physical ability investigated with the 6MWT at T_1 and T_2 [25,37-39]. The choice of the 6MWT rather than the VO_2max as a measure of physical ability is because of the multicentric real-life study design.

The secondary outcome parameters are: (1) fatigue, the leading symptom of sarcoidosis, investigated using the FAS at T_1 to T_4 [28,31,40-43]; (2) the psychological burden assessed with the HADS-D at T_1 to T_4 [26]; (3) HRQL assessed by the generic SF-36 health survey questionnaire at T_1 to T_4 [27,44-47]; (4) the compiled patients' views of: the components of the individual rehabilitation programs and their perceived importance at T_1; and (5) an evaluation of the program from the patients' perspective at T_1.

To achieve a 1-year follow-up of patients who return to their homes all over Germany after the end of the inpatient rehabilitation, after T1 validated questionnaires sent by post were chosen as the only data source. Dependent on the outcome of the study, a repeat of the objective measurements (lung function and exercise test) after 3, 6, or 12 months is planned as part of a RCT follow-up study.

Data Management and Statistical Analysis Procedures

Data management will be carried out by the Centre for Public Healthcare and statistical analysis by the Institute for Biostatistics at the Hannover Medical School.

The questionnaires will be anonymized using a clinic identifier and a consecutive patient number and will be digitized in duplicate at the Hannover Medical School and entered in Microsoft Access. The database tables will then be virtually superposed to identify differences between entries. When discrepancies are found, the data entry will be compared with that of the original questionnaire and corrected so that an accurate master dataset used for the descriptive analysis and the calculation of effect sizes will emerge.

The main analyses will be performed on the total study population. Missing values will be reported and not replaced initially. Changes in both primary endpoints between T_0 and T_1 will be evaluated with a paired *t* test and a 2-sided type I error of 2.5%.

Because ProKaSaRe is an exploratory study, for all analyses except for the primary analysis, *P* values will be assessed descriptively and considered to be of importance for $P<.05$. Therefore, no adjustment for multiplicity will be performed. Descriptive evaluation of clinical characteristics will be conducted. Changes from baseline for continuous data will be compared by paired *t* tests and given with 95% confidence intervals for the mean difference. Categorical parameters will be compared for T_1 to T_4 by chi-square test. Duration of sick leave and results of the sociomedical status documented by the physician (T_0, T_1) and by the patient (T_2, T_3, T_4), the evaluation of rehabilitation from the patients´ perspective (T_1) and patient-attributed value to the components of the individual rehabilitation program will be presented exploratively. For the questionnaires, a covariate analysis will be run using participating centers and baseline values as independent variables and the change from T_1 and T_2 to T_0 as dependent variables. Further multivariate analyses are planned.

All statistical analyses will be conducted using the SAS 9.3 software (SAS Institute Inc, Cary, NC, USA) to show the impact of the rehabilitation program on the patients' lives.

Data Quality Check

The planned double data entry and the possibility of referring to the clinical files in case of runaway values and spikes concerning lung function, gas exchange, and laboratory parameters should ensure high data quality.

Intervention

The intervention is an inpatient rehabilitation program lasting 3 weeks. The program is modular, with obligatory and optional components. All components are available at all study centers (Textbox 1). The reasons for choosing these interventions rather than others is described briefly in the following.

Rehabilitation is oriented according to the nature and extent of the difficulties resulting from single (sarcoidosis) or multiple illnesses (comorbidities) that the individual patient suffers that affect professional and daily life. According to these disabilities and functional deficiencies (as defined by the rehabilitation admission tests and assessments), the therapy components are individually and quantitatively adapted.

Textbox 1. Components of the (usual) rehabilitation program carried out by all participating sites in ProKaSaRe.

Obligatory components

- Specialist confirmation of diagnosis and checks of current medication.
- Endurance training: from 3 to 5 training sessions per week at approximately 60-80% of maximum capacity (walking, power walking, or cycle ergometer).
- Strength training: 3 medical training therapy sessions per week.
- Respiratory physiotherapy: at least three group sessions per week and at least two individual respiratory therapy sessions during the rehabilitation program.
- Patient education: modular sarcoidosis education program.

Optional components

- Inspiratory muscle training: at least five sessions (each 30 minutes) per week, starting at 30% PI_{max}. Sessions supervised at least once a week.
- Relaxation treatment: autogenic training or progressive muscle relaxation.
- Social counseling.
- Nutrition counseling.
- Psychological individual and group therapy.
- Structured smoking cessation.
- Ergotherapy and advice on devices and equipment.
- Patient education: additional courses (eg, long-term oxygen therapy training).

Obligatory Components

Endurance training is intended to improve performance, shortness of breath, mood, and quality of life. It also compensates for cortisone adverse effects, such as osteoporosis, muscle degeneration, weight gain, diabetes, and high blood pressure [15,48,49], allowing improvement in activities of daily life as defined by the International Classification of Functioning, Disability and Health (ICF) [50]. Cortisone may also affect the mood, sometimes toward depression or irritability.

Respiratory physiotherapy supports the learning of coughing techniques, positions which make breathing easier, handling shortness of breath on exertion, stretching positions to maintain the flexibility of the bony thorax (particularly with fibrosis), correct breathing techniques, and improvements in the respiratory muscles.

Patient education provides the sick person with information on the clinical picture, helps them to cope with their illness, and deal with all the information on adverse effects of their therapies (eg, cortisone). It puts sarcoidosis into perspective and helps to reduce anxiety and depression by generating feelings of informed safety. It explains the reasons for and frequencies of check-ups and helps patients to differentiate between required and unnecessary diagnostic tests. Alternative therapies and their evaluation with respect to sarcoidosis treatment could be addressed. Finally, it generates opportunities for strong interaction and bonding with other patients.

Optional Components

Inspiratory muscle training is known to improve the strength of respiratory muscles [51]. Relaxation treatment helps to reduce the symptoms of disquiet, stress, and anxiety.

Social counseling provides advice relating to societal participation, level of disability, severe disability, career reintegration according to social statutes and the ICF, taking early retirement, and applying for benefits, etc [49,52,53].

Nutrition counseling will also be offered on demand, particularly with reference to the adverse effects of cortisone therapy, such as osteoporosis, increased weight, diabetes, and high blood pressure, including counseling on the subject of a healthy diet.

Psychological individual and group therapy will be offered when needed. Individual therapy includes counseling and advice when anxiety, depression, insecurity, and problems coming to terms with the illness occur due to the chronic nature of the illness [54]. Structured smoking cessation will be offered to all smokers. Ergotherapy and advice on devices and equipment will be occasionally appropriate as part of physiotherapy.

Results

Funding was obtained on October 12, 2010; enrollment began on January 15, 2011 and was completed by January 14, 2012. Results are anticipated late summer 2015.

Discussion

Sarcoidosis is a systemic disease of unknown etiology that can affect all organs. Therefore, it presents a heterogeneous clinical picture associated with multiple comorbidities. It can occur at any age with a peak between the ages of 20 and 40, with women having a higher frequency of illness than men do, and it can show a chronic or chronic-relapsing pattern. Significant functional limitations include reduced quality of life and an often-debilitating fatigue that hinders the patients' participation in "normal" daily life. Sarcoidosis is often insufficiently treatable by drugs alone. Can rehabilitation slow down disease progression and accelerate recovery and return to employment? ProKaSaRe seeks to find out whether and to what extent a rehabilitative intervention can substantially contribute to the care of patients with sarcoidosis.

This paper outlines the rationale and design for a prospective, "real-life" multicenter study using obligatory and optional components administered by a multiprofessional team. The aim is to explore the effects of a 3-week inpatient multiprofessional pulmonary rehabilitation program. ProKaSaRe will supply lacking evidence as to the degree to which a training-based multimodal rehabilitation can produce or enhance a significant and sustained improvement to HRQL, physical fitness, and psychological distress. This is particularly relevant in light of the myasthenia and fatigue mentioned previously, which are common symptoms of sarcoidosis.

During 2011, 929 sarcoidosis patients were admitted for inpatient rehabilitation in Germany [55]. We plan to enroll at least 121 consecutive patients within ProKaSaRe, a number that will encompass almost a seventh of all patients undergoing rehabilitation for sarcoidosis in Germany during the year of the study. By using a recruitment time of 12 months, seasonal influences will be excluded, which adds to the validity of the findings.

ProKaSaRe will help to define the patient group currently being referred for pulmonary rehabilitation in Germany, while simultaneously documenting the effect of pulmonary rehabilitation in terms of clinically relevant outcome parameters, if any.

The inpatient rehabilitation that will be assessed consists of a clearly defined set of components developed and approved by experts in the field accredited by the German health system following assessment of their professional development and experience as heads of specialized centers. The detailed agreed-upon choice of components and the broad assessment of HRQL reflecting the subsequent patients' reaction to their chosen and advised measures represent the strengths of ProKaSaRe.

Scientists and physicians tend to define therapeutic success by only the improvement in pulmonary function, whereas patients equate successful treatment with tangible effects in their daily lives and an improved sense of well-being. This latter ultimately determines long-lasting positive effects, as stated by the ICF [50]. For this reason, both measurements (clinical data and patients' self-evaluation) are necessary to form a balanced picture of pulmonary rehabilitation outcomes.

Although ProKaSaRe could contain some bias because it assesses only the sarcoidosis population willing to undergo pulmonary rehabilitation and is not a RCT, it is nevertheless a

real-life study. Therefore, it is likely that the results generated in ProKaSaRe will be highly generalizable.

In contrast to studies focusing on single nonpharmaceutical components of rehabilitation programs, ProKaSaRe considers the entirety of the pulmonary rehabilitation program. The participants represent a real-life sample of sarcoidosis patients rather than a selected subpopulation thereby adding to the importance of the findings for medical and economical deliberations.

If the results of ProKaSaRe show effectiveness of the rehabilitation over a long period of time, rehabilitation as documented by the study would offer perspective and justified hope to concerned patients and their families and also evidence to stakeholders in the health care system. The documented efficacy of pulmonary rehabilitation for sarcoidosis patients will fill a gap in the reference literature used to develop guidelines and efficient long-lasting treatment procedures. It may establish rehabilitation as an irreplaceable and necessary additional therapeutic option for sarcoidosis, with highly relevant outcomes, which is only effective in its entirety and is more than the sum of single administrated components.

Depending on the outcome of ProKaSaRe, an RCT will be planned to advance knowledge and funding will be applied for. The definition of patient groups to which rehabilitation offers the greatest benefit and the estimation of the financial savings by the prescription of a rehabilitation program would be the next topics to be investigated.

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Authors' Contributions

HL designed the data collection tools, monitored data collection for the whole study, wrote the statistical analysis plan, cleaned and analyzed the data, and drafted and revised the paper. AG wrote the statistical analysis plan and revised the draft paper. KF analyzed the data and revised the paper. HBS monitored data collection for the whole study and revised the draft paper JvdM monitored data collection for the whole study and revised the draft paper. UT monitored data collection for the whole study and revised the draft paper. RH monitored data collection for the whole study and revised the draft paper. KS initiated and coordinated the collaborative study, designed data collection tools, monitored data collection for the whole study, and drafted and revised the paper.

Conflicts of Interest

None declared.

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Abbreviations

6MWT: 6-minute walk test
ACE: angiotensin-converting enzyme
BMI: body mass index
COPD: chronic obstructive pulmonary disease

CRP: C-reactive protein
FAS: Fatigue Assessment Scale
FEV1: forced expiratory volume in 1 second
HADS-D: Hospital Anxiety and Depression Scale
HRQL: health-related quality of life
ICF: International Classification of Functioning, Disability and Health
mMRC: Modified Medical Research Council Dyspnea Scale
Pimax: maximal inspiratory mouth pressure
P0.1: mouth occlusion pressure
RCT: randomized controlled trial
RV: residual volume
SF-36: 36-item Short Form Health Survey
SGRQ: St George's Respiratory Questionnaire
sRaw: specific airway resistance
TLC: total lung capacity
Tlco: carbon monoxide transfer factor
Tlco SB: carbon monoxide transfer factor in a single breath
Tlco/Va: carbon monoxide transfer factor per alveolar volume
VC: vital capacity

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Original Paper

Detecting Heart Failure Decompensation by Measuring Transthoracic Bioimpedance in the Outpatient Setting: Rationale and Design of the SENTINEL-HF Study

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Abstract

Background: Recurrent hospital admissions are common among patients admitted for acute decompensated heart failure (ADHF), but identification of patients at risk for rehospitalization remains challenging. Contemporary heart failure (HF) management programs have shown modest ability to reduce readmissions, partly because they monitor signs or symptoms of HF worsening that appear late during decompensation. Detecting early stages of HF decompensation might allow for immediate application of effective HF therapies, thereby potentially reducing HF readmissions. One of the earliest indicators of HF decompensation is intrathoracic fluid accumulation, which can be assessed using transthoracic bioimpedance.

Objective: The SENTINEL-HF study is a prospective observational study designed to test a novel, wearable HF monitoring system as a predictor of HF decompensation among patients discharged after hospitalization for ADHF.

Methods: SENTINEL-HF tests the hypothesis that a decline in transthoracic bioimpedance, as assessed daily with the Philips fluid accumulation vest (FAV) and transmitted using a mobile phone, is associated with HF worsening and rehospitalization. According to pre-specified power calculations, 180 patients admitted with ADHF are enrolled. Participants transmit daily self-assessments using the FAV-mobile phone dyad for 45 days post-discharge. The primary predictor is the deviation of transthoracic bioimpedance for 3 consecutive days from a patient-specific normal variability range. The ADHF detection algorithm is evaluated in relation with a composite outcome of HF readmission, diuretic up-titration, and self-reported HF worsening (Kansas City Cardiomyopathy Questionnaire) during a 90-day follow-up period. Here, we provide the details and rationale of SENTINEL-HF.

Results: Enrollment in the SENTINEL-HF study is complete and the 90-days follow-up is currently under way. Once data collection is complete, the study dataset will be used to evaluate our ADHF detection algorithm and the results submitted for publication.

Conclusion: SENTINEL-HF emerged from our long-term vision that advanced home monitoring technology can improve the management of chronic HF by extending clinical care into patients' homes. Monitoring transthoracic bioimpedance with the FAV may identify patients at risk of recurrent HF decompensation and enable timely preventive measures.

Trial Registration: Clinicaltrials.gov NCT01877369: https://clinicaltrials.gov/ct2/show/NCT01877369 (Archived by WebCite at http://www.webcitation.org/6bDYl0dGy)

(JMIR Res Protoc 2015;4(4):e121) doi:10.2196/resprot.4899

KEYWORDS

acute decompensated heart failure; rehospitalization; remote monitoring; transthoracic bioimpedance; wearable fluid accumulation vest

Introduction

Hospitalizations related to acute decompensated heart failure (ADHF) have increased over recent decades [1], and ADHF is now the leading cause of hospital admissions in elderly patients [2]. In light of the high cost related to inpatient treatment of ADHF [3], there is increasing interest in deploying telehealth technologies to extend clinical care into the homes of patients with heart failure (HF), thereby allowing for more frequent assessment for and prevention of HF decompensations. However, the identification of HF patients in the early stages of decompensation remains a major clinical challenge. Significant efforts have focused on developing HF management programs to identify and intervene upon patients with or at-risk for ADHF in order to decrease HF-related hospitalizations [4-7]. Contemporary HF management programs rely on general, medication-related, and disease-specific patient education as well as active surveillance for signs or symptoms of acute HF decompensation. The impact of such programs has been limited, in part due to their cost, but also because HF surveillance tools, including measurements of heart rate, blood pressure, and body weight, show only modest abilities to identify individuals at risk for HF decompensation. Developing a home-based monitoring system using markers that can detect ADHF in its early stages is not only acceptable to elderly patients and facilitates communication between clinicians and patients, but has the potential to reduce rates of HF decompensation and hospitalization.

Novel measures of bioimpedance, or opposition to electric current through body tissues, can be used to identify fluid accumulation [8]. Prior studies have demonstrated that intrathoracic bioimpedance, measured in HF patients with implantable cardioverter-defibrillators, is a valid predictor of clinical events including ADHF and hospitalization [9]. Nevertheless, few patients with HF meet criteria for implantation of implantable cardioverter-defibrillators. More generalizable and less invasive methods to measure bioimpedance would capture a more representative population of patients with HF. Early data suggest that transthoracic bioimpedance measured using a wearable, investigational device may also detect intra-thoracic volume retention [10,11] and correlate with intrathoracic measures [12].

In order to explore the hypothesis that transthoracic bioimpedance assessed daily with a novel, non-invasive, wearable fluid accumulation vest (FAV) and transmitted using a mobile phone would identify patients at risk for HF related rehospitalization, we designed SENTINEL-HF, a prospective study of patients discharged after hospitalization for ADHF. Here, we describe the design and rationale of SENTINEL-HF.

Methods

Device Description

The FAV is a non-invasive wearable monitor designed to spot-check transthoracic bioimpedance and send the measurement data to a remote telehealth center. The FAV system consists of a measurement vest, an electronics module, a mobile phone-based app, and a remote database (Figure 1). The measurement vest is a functional textile that is fitted snugly to the patient's chest circumference using an adjustment strap and enables at-home self-measurements [13]. The inside of the vest has 4 textile electrodes arranged pair wise on 2 supporting pads located on either side of the rib cage, at the inferior part of the lungs. The electronics module connects to the back of the vest. The module determines transthoracic bioimpedance at multiple frequencies, which enables a model-based assessment of intra- and extra-cellular fluid status within the thorax [14] and the detection of respiration [15]. The module also records a 1-lead ECG as well as the patient's motion and posture. The electronics module communicates wirelessly with a mobile phone-based app that guides the user through the measurement steps, controls the measurement parameters, and receives the measurement data. After each measurement, the app automatically transmits the data to a remote database hosted by the sponsor in a secure data center.

Self-assessments with the FAV are performed by following a simple measurement routine that takes 8-10 minutes (Figure 2).

Figure 1. Components of the fluid accumulation vest (FAV) measurement system and the process for data acquisition, transfer, and storage.

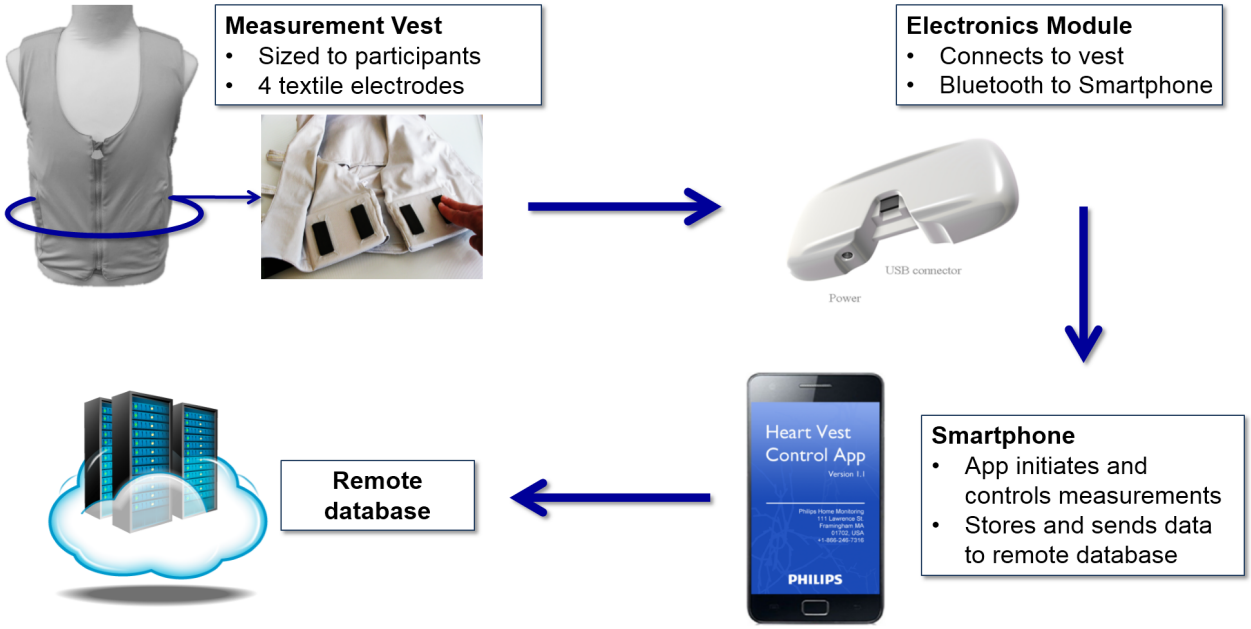
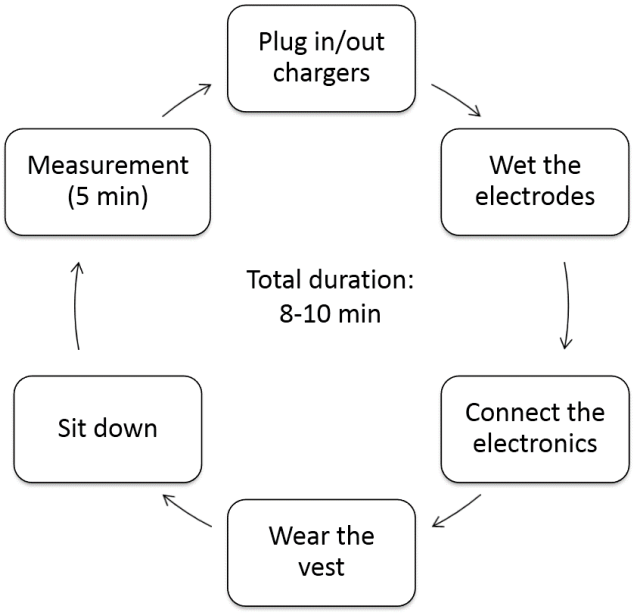


Figure 2. The daily measurement routine consists of wetting the electrodes (eg, using wetted fingertips), connecting the electronics module to the measurement vest, wearing the vest, sitting down, initiating a measurement with the mobile phone-based app, remaining seated for 5 minutes while the measurement is in progress, taking the vest off, and docking the devices for charging until the next day's measurement.



Aim and Design

The aim of the SENTINEL-HF study is to evaluate whether transthoracic bioimpedance assessed daily using the FAV system following discharge from an HF hospitalization can predict recurrent HF decompensation events. The study also aims to assess patient adherence to the FAV system and delineate adherence profiles based on novel patient demographic and psychosocial characteristics.

SENTINEL-HF is a prospective, non-randomized, observational study. Patients are enrolled during a baseline hospitalization for HF and are monitored for 45-days post-discharge by means of daily self-assessments with the FAV. Patient-reported data related to healthcare utilization and quality of life are collected through interviews conducted during the baseline hospitalization and throughout the home monitoring period, at days 7, 14, and 45. Passive surveillance of participants for clinical events is performed through medical record review for 90 days following discharge. The extended passive surveillance period enables the examination of bioimpedance trends in relation with long-term outcomes.

Approximately 180 patients are enrolled at 2 teaching hospitals that comprise the University of Massachusetts Memorial Medical Center (UMMMC), a large academic medical center in Central Massachusetts. The UMMMC serves a heterogeneous patient population with a diverse ethnic/racial, socioeconomic,

urban/rural, and other socio-demographic background. Medicare 30-days readmission and death rates for patients discharged from an HF-related hospitalization at UMMMC are similar to the US national rates (23% and 12%, respectively) [16].

Study Population

The SENTINEL-HF study enrolls patients in New York Heart Association (NYHA) functional classes II-IV who are hospitalized with a primary diagnosis of HF or with a sign (ie, vascular congestion on chest radiograph, rales on lung exam, or peripheral edema) and a symptom (ie, dyspnea or orthopnea) consistent with ADHF. Adult patients (≥21 years) are eligible to participate if they are willing and able to comply with the clinical investigation plan, in particular, able to handle the technical devices, and are available for follow-up visits throughout a 45-days post-discharge period. Exclusion criteria include an N-terminal pro-brain natriuretic peptide (NT-proBNP) level ≤100 pg/dL, end-stage chronic kidney disease requiring hemodialysis, chronic obstructive pulmonary disease (COPD), a primary diagnosis of gastrointestinal bleed, asthma, cardiac arrest or acute coronary syndrome at baseline, primary pulmonary hypertension, psychiatric or neurological disorders of moderate to severe degree, non-English speakers, pregnant women, prisoners, patients planning to move from their residence within 2 months from baseline, a body habitus that prevents the patient from fitting into a measurement vest or does not allow for adequate electrode-skin contact while wearing the vest, non-intact skin at the location of the electrodes, or the presence of an implantable pacemaker/defibrillator.

Endpoints

The ability of the FAV system to predict HF decompensation events is evaluated in relation to the composite endpoint of unplanned HF-related rehospitalization and HF worsening. A rehospitalization is defined as a presentation to the emergency department or any unplanned admission into a hospital environment with at least one overnight stay. HF worsening is defined as either diuretic up-titration or HF-related worsening quality of life, as assessed by the Kansas City Cardiomyopathy Questionnaire [17]. Secondary endpoints of the study include major adverse cardiac events, emergency department visits, all-cause rehospitalization, and death.

Study Procedures

SENTINEL-HF's rich data collection is focused on 3 main activities (home monitoring, interviews, and event tracking), which are initiated during the baseline hospitalization and continued up to 90-days post-discharge. The study procedures are summarized in Figure 3 and are detailed in the following sections. Data collected during the study is outlined in Table 1.

Figure 3. Diagram of the SENTINEL-HF study. This is a prospective, observational study in patients discharged after a baseline hospitalization for acute decompensated heart failure (ADHF). Participants will be recruited during the baseline hospitalization and follow-up activities will be initiated before discharge. Follow-up consists of 3 pillars: home monitoring which includes a period of self-assessments with the fluid accumulation vest (FAV), participant interviews, and event tracking based on participants' electronic medical records (EMR).

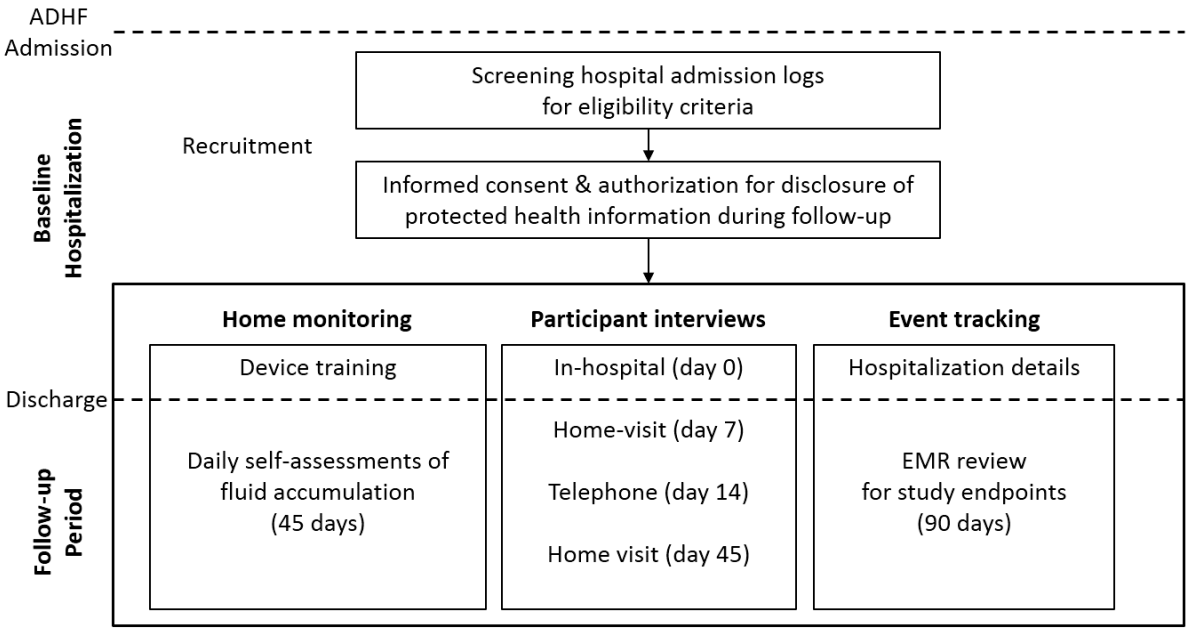


Table 1. Data collected in SENTINEL-HF.

Domains and measures		Date of collection				
		Baseline	Day 7	Day 14	Day 45	Day 90
Demographics						
	Date of birth	X				
	Gender	X				
	Marital status	X				
	Race/ethnicity	X				
	Height	X				
	Weight	X				
	Thorax circumference	X				
Baseline hospitalization details						
	Chief complaint at admission	X				
	ICD-9 codes of hospitalization (first 3)	X				
	Medical histories and comorbidities	X				
	Type of cardiac dysfunction	X				
	Type of ventricular failure	X				
Clinical events and outcomes						
	Readmissions		X	X	X	X
	Emergency room visits		X	X	X	X
	Outpatient encounters		X	X	X	X
	Medication changes	X	X	X	X	X
Patient reported outcomes: transition quality						
	Care Transitions CTM-319		X	X	X	
Patient reported outcomes: quality of life						
	SF-12v2™ Health Survey [18]	X	X		X	
	Kansas City Cardiomyopathy Questionnaire (KCCQ) [17]	X	X	X	X	
	Self-Care of Heart Failure Index [19]	X	X	X	X	
Psychosocial characteristics						
	Capacity of informed consent [20]	X				
	Telephone interview for cognitive status (TICS) [21]	X	X	X	X	
	Patient Health Questionnaire (PHQ-9) [22]	X	X		X	

Baseline Procedures

Patients with possible ADHF are identified using a computerized patient tracking system in the emergency department and by monitoring hospital admission logs on a daily basis. Screening is performed based on the diagnosis, chief complaint, and medical history. The eligibility of potential participants is confirmed based on their medical record information including laboratory records, ECG reports, and physical examination findings. Eligible patients are approached during hospitalization and provided with information about the study. Patients who agree to participate sign an informed consent and an authorization for disclosure of protected health information during the follow-up period (eg, details of subsequent readmissions). If study staff have concerns about the capacity of a potential participant to provide informed consent, an assessment [20] is conducted to determine the patient's ability to express choice, to understand relevant information, and to appreciate the situation and its likely consequences. Patients who fail the assessment instrument are ineligible for the study.

Shortly before discharge, each enrolled participant is provided with a personal FAV monitoring kit consisting of a measurement vest of appropriate size, an electronics module, a study dedicated

mobile phone, a manual of operation, and a short form instructions and troubleshooting guide. Staff adjust the measurement vest to the participant's chest circumference ensuring that the electrodes have good contact with the participant's skin, and train participants, and their caregivers when applicable, to use the study equipment.

Home Monitoring

During a 45-day home monitoring period, participants are required to perform daily self-assessments with the FAV at a consistent time of day, preferably upon waking up in the morning. Study staff use scheduled follow-ups to ensure that the self-assessments are carried out correctly and, if necessary, to provide assistance in the form of additional instructions for using the FAV. On day 1, staff call each enrolled participant to check on their experience with the first FAV self-assessment. On day 7, study staff visit the participants at home providing a booster training for the use of the FAV system. On day 14, study staff call the participants encouraging them to continue with the daily self-assessments throughout the following month (Figure 3). Participants who require additional assistance at any time during the home monitoring period may call a study hotline maintained by the sponsor (Philips Healthcare).

Participants' adherence to the daily self-assessment routine is monitored based on the data transmitted to the remote database. Study staff call participants with 2 consecutive missed or non-evaluable days to identify problems and to provide support in resuming the daily self-assessments. If 2 consecutive missed or non-evaluable days continue to occur after the call, study staff visit the struggling participants at home to troubleshoot technical issues and to walk the participants through the FAV self-assessment routine. Participants are removed from the study if 2 consecutive missed or non-evaluable days continue to occur after the home visit.

Participant Interviews

In order to identify characteristics associated with the acceptance and the successful use of the FAV, participant interviews are conducted in-person or over the telephone at baseline and at day 7, 14, and 45 post-discharge (Figure 3). During these follow-up interactions, key patient reported outcomes and covariates, including severity of depressive symptoms and HF self-care are assessed. An overview of collected data and instruments used in the interviews is provided in Table 1.

At each follow-up interview, hospital-to-home transition quality is assessed using the 3-item Care Transition Measure (CTM3), an assessment designed to measure the quality of care transitions from the patient's perspective [23]. The measure has good psychometric properties and has predictive validity for rehospitalizations. We also assess for HF self-management, patient response to changes in symptoms of HF, and confidence with self-management using the validated Self-Care for Heart Failure Index [19]. We measure both general and disease-specific quality of life at baseline and during each follow-up interview. General quality of life is measured by using the Short Form Health Survey SF-12, a validated 12-item version of the SF-36, a quality of life instrument often used for patients with heart disease [18]. HF-specific quality of life is assessed using the Kansas City Cardiomyopathy Questionnaire (KCCQ) [17].

Each participant's psychosocial profile is characterized for a better understanding of factors associated with successful FAV use. Cognitive impairment and depressive symptoms are assessed at baseline and during each follow-up interview using the telephone interview for cognitive status (TICS) [21] and the 9-item Patient Health Questionnaire (PHQ-9) [22], respectively. We also gather information about clinical endpoints, including medication changes, doctor's visits, emergency department visits, or readmissions. These reports are confirmed and extended by reviewing each participant's medical record.

Baseline Clinical Characteristics and Endpoint Review

Clinical information (medical history, physical exam data, chest x-ray findings, and serum laboratory values) and demographics (age, gender, marital status, race/ethnicity, height, weight, and blood pressure and heart rate) are abstracted at baseline from the participants' electronic medical records. Surveillance of participants continues for 90 days post-discharge by reviewing all additional clinical data including hospitalizations or physician visits related to possible ADHF, cardiac studies, consultations, discharge summaries, emergency service records, lab reports, office and clinic notes, operative and procedure reports, problem lists, pulmonary studies, and radiology results from the electronic medical records. Because the majority (80-90%) of patients hospitalized for ADHF at UMMMC follow-up with a UMMMC cardiologist and are rehospitalized at UMMMC, the vast majority of participant data is available in one centralized database of electronic medical records. Information related to patient-reported clinical events occurring outside the UMMMC health system is obtained from the respective healthcare provider based on the participant's consent for solicitation of outside records. Endpoints are adjudicated on a rolling basis by 3 expert physicians (authors TEM, DDM, CD) blinded to FAV measures and clinical outcomes.

Data Analysis

The study database is used to evaluate the performance of a previously developed ADHF detection algorithm informed by bioimpedance measures.

Parameter Extraction

Relevant tissue characteristics are extracted from each FAV self-assessment by fitting the Cole-Cole model (Figure 4) to the multifrequency bioimpedance data collected by the device [14,24]. The model parameters reflect extracellular fluids (R_E), intracellular fluids (R_∞), tissue relaxation (fc), and tissue heterogeneity (α). Thoracic fluid accumulation due to worsening HF directly affects the value of R_E which is expected to decrease [8,11]. Therefore, R_E will be used as the primary bioimpedance index to predict HF decompensation.

Figure 4. Cole-Cole model equation.

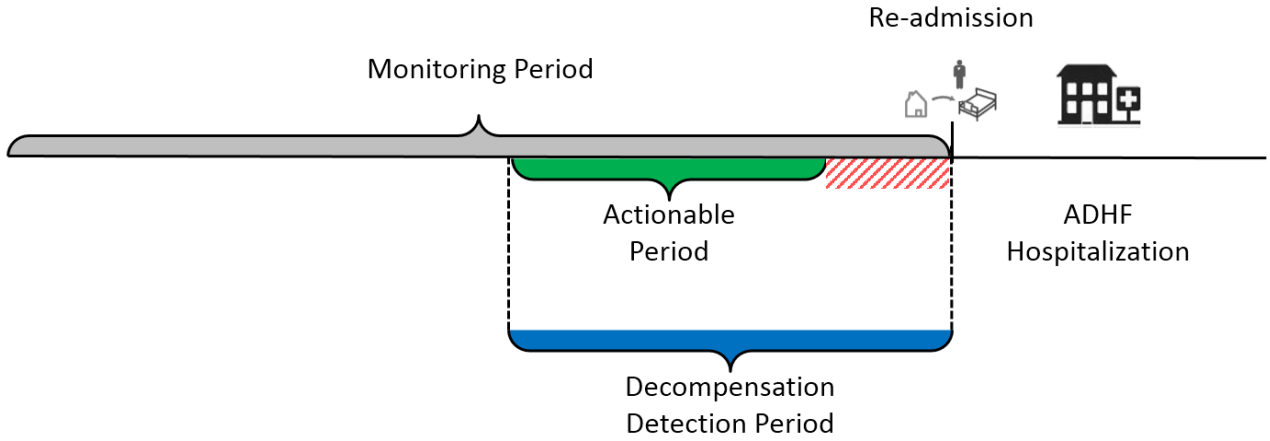
$$Z = R_\infty - \frac{R_\infty - R_E}{1 + \left(j\frac{f}{f_c}\right)^\alpha}$$

Acute Decompensated Heart Failure (ADHF) Detection

An ADHF detection algorithm was designed to monitor the daily evolution of a patient's bioimpedance index. The algorithm determines if a new value is normal by comparing it to a patient-specific range of normal variability. The occurrence of abnormal values of the bioimpedance index causes the algorithm to raise decompensation alerts. In a real-life use case, decompensation alerts have the potential to prevent HF events, for example hospitalizations, only if they are raised a sufficient number of days prior to an acute decompensation allowing sufficient time for healthcare providers to intervene and for administered therapies to take effect. Because telehealth alerts are generally reviewed during business hours on weekdays, the reaction to an alert may be delayed by 2-3 days if an alert is raised at the beginning of a weekend. To accommodate for this in the evaluation of the ADHF detection algorithm we define the actionable period (AP) as the time window preceding an endpoint in which decompensation alerts are most likely to contribute to an improved patient outcome. The algorithm is evaluated based on its ability to raise alerts within the AP and any alert raised after the AP is excluded from the performance evaluation (Figure 5). The length and the end of the AP can be set by each healthcare provider individually. While healthcare providers generally agree that a realistic length for the AP is likely to be shorter than 30 days, setting the end of the AP remains a topic of debate. Therefore, we set the boundaries of the AP based on the performance achieved by current standard of care telehealth systems which include measurements of weight, heart rate, or blood pressure. These systems typically use weight as the primary predictor of decompensation and have the ability to raise decompensation alerts 2-3 days prior to an impending HF-related hospitalization [25]. In the analysis of the SENTINEL-HF study, we evaluate whether the FAV is able to extend this period by enabling earlier decompensation alerts. To this aim, we vary the end of AP down to 2 days prior to an endpoint event and the length of the AP between 10 and 30 days.

The predictive accuracy of the ADHF detection algorithm is assessed based on the number of true positive, true negative, false positive and false negative alerts preceding an endpoint. Prediction performance metrics are assessed in relation with the primary and the secondary endpoints, respectively.

Figure 5. The ADHF detection strategy. Example of an ADHF hospitalization occurring during the FAV monitoring period. The ADHF detection algorithm is evaluated based on its ability to raise decompensation alerts within an actionable period which ends days prior to the event.



Sample Size Calculations

Our prospective sample size calculations were guided by the a priori assumption that the FAV would have a sensitivity of 0.75 to detect a primary endpoint of ADHF hospitalization. This assumption was based on a previous dataset collected for the development of our ADHF detection algorithm [12]. In SENTINEL-HF, we plan to have 107 patients complete the study period and adhere to the FAV daily over 45-days to exclude a real sensitivity lower than 0.60. To account for non-evaluable data and a 30% dropout rate, we require a minimum of 160 participants to provide a 0.1 95% confidence interval around an observed sensitivity of 0.75. We are employing an adaptive study design with interim sample size re-assessments based on the observed drop-out rate, non-evaluable data, and the number of ADHF events.

Ethical Conduct and Study Management

The SENTINEL-HF study is conducted in compliance with Good Clinical Practice and the principles outlined by the Declaration of Helsinki. The study has been approved by the Committee for the Protection of Human Subjects at University of Massachusetts Medical School with Institutional Review Board number H00001760. All study participants provide written informed consent at enrollment. The study is sponsored by Philips Healthcare and is monitored by the sponsor in compliance with the Code of Federal Regulations. Unanticipated adverse device effects are reported to the sponsor and to the

Massachusetts Medical School Institutional Review Board. The study trial registration number is NCT01877369.

Results

SENTINEL-HF enrolled the first patient in June 2013 and the last patient in April 2015. A total of 180 patients are enrolled in the study. Once the 90-days follow-up of the last patient is complete, the SENTINEL-HF dataset will be used to evaluate our ADHF detection algorithm and first results will be submitted for publication.

Discussion

The SENTINEL-HF study is a prospective, non-randomized, observational study evaluating the ability of a wearable HF monitor to anticipate ADHF events. Patients perform daily self-assessments of transthoracic bioimpedance for 45 days following discharge from a HF hospitalization. Patient-reported quality of life are assessed using validated questionnaires and clinical events are abstracted from the patients' electronic medical records. Endpoints adjudicated by a team of physicians are used to evaluate the performance of an ADHF detection algorithm informed by serial bioimpedance measurements. SENTINEL-HF supports our long term vision that advanced home monitoring technology can aid the management of chronic HF by extending clinical care into patients' homes, thereby preventing HF decompensations and reducing hospitalizations.

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Conflicts of Interest

The SENTINEL-HF study is sponsored by Philips Healthcare (Andover, US). SD and JR are employees of Philips Research, and FSK is an employee of Philips Healthcare.

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Abbreviations

ADHF: acute decompensated heart failure
AP: actionable period
FAV: fluid accumulation vest
HF: heart failure
UMMMC: University of Massachusetts Memorial Medical Center

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Original Paper

Using an eHealth Intervention to Stimulate Health Behavior for the Prevention of Cognitive Decline in Dutch Adults: A Study Protocol for the Brain Aging Monitor

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Abstract

Background: Internet-delivered intervention programs are an effective way of changing health behavior in an aging population. The same population has an increasing number of people with cognitive decline or cognitive impairments. Modifiable lifestyle risk factors such as physical activity, nutrition, smoking, alcohol consumption, sleep, and stress all influence the probability of developing neurodegenerative diseases such as Alzheimer's disease.

Objective: This study aims to answer two questions: (1) Is the use of a self-motivated, complex eHealth intervention effective in changing multiple health behaviors related to cognitive aging in Dutch adults in the work force, especially those aged 40 and over? and (2) Does this health behavior change result in healthier cognitive aging patterns and contribute to preventing or delaying future onset of neurodegenerative syndromes?

Methods: The Brain Aging Monitor study uses a quasi-experimental 2-year pre-posttest design. The Brain Aging Monitor is an online, self-motivated lifestyle intervention program. Recruitment is done both in medium to large organizations and in the Dutch general population over the age of 40. The main outcome measure is the relationship between lifestyle change and cognitive aging. The program uses different strategies and modalities such as Web content, email, online newsletters, and online games to aid its users in behavior change. To build self-regulatory skills, the Brain Aging Monitor offers its users goal-setting activities, skill-building activities, and self-monitoring.

Results: Study results are expected to be published in early 2016.

Conclusions: This study will add to the body of evidence on the effectiveness of eHealth intervention programs with the combined use of state-of-the-art applied games and established behavior change techniques. This will lead to new insights on how to use behavior change techniques and theory in multidimensional lifestyle eHealth research, and how these techniques and theories apply when they are used in a setting where no professional back-end is available.

Trial Registration: Nederlands Trial Register: NTR4144; http://www.trialregister.nl/trialreg/admin/rctview.asp?TC=4144 (Archived by WebCite at http://www.webcitation.org/6cZzwZSg3)

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KEYWORDS

cognition; healthy lifestyle; eHealth; internet; prevention; applied games; protocol

Introduction

Multiple systematic reviews and meta-analyses have shown that Internet-delivered intervention programs aimed at health behavior change can have a positive impact on their respective populations. These effects range from weight loss in obese men and women to moderating alcohol intake patterns, smoking cessation, and adjusting dietary patterns [1-4]. Computer-tailored health programs are complex, long-term programs that are appropriate for targeting multiple behaviors requiring a change in behavioral habits [3,5]. In contrast to results reported by Portnoy et al who showed that increasing age was a negative predictor of program effectiveness, a systematic review by our group on the effectiveness of eHealth interventions in older populations provides evidence that older age cohorts can be reached with eHealth interventions [6]. Within the next few years, the upcoming cohort of older adults will be adapted to a new electronic environment, in contrast to previous cohorts who often lacked computer experience and had limited Internet access. This would facilitate further use of eHealth tools, also for prevention targets in the elderly. With Internet penetration in the Netherlands reaching 94% of all households among the population aged 45-75, more widespread use of eHealth by the elderly is likely.

We use Bennett et al's definition for Internet interventions as "systematic treatment/prevention programs, usually addressing one or more determinants of health (frequent health behaviors), delivered largely via the Internet (although not necessarily exclusively Web-based), and interfacing with an end user" [7]. These eHealth interventions are characterized by being highly structured, mostly self-guided, interactive, visually rich, and they may provide tailored messaging based on end-user data [2,8]. Additional benefits of Internet programs are their 24-hour availability, uniformity in data dissemination and collection [2,9], and their heightened reach [5]. An advantage of creating such a completely self-motivated eHealth program, in comparison to an expert-led intervention, is the fact that the reduction in needed external support exponentially increases the reach of Internet interventions. Thus, Internet interventions can reach as many participants as is technically allowed by the hosting servers [10]. Moreover, since most of the cost of Web-delivered health programs are associated with the development stages rather than the implementation stage itself (in comparison to regular face-to-face treatment), even programs with relatively low effectiveness but a very large reach, could significantly impact public health [4].

Lifestyle interventions through the Internet are not new. However, online lifestyle intervention programs with cognitive functioning as a primary outcome measure are not yet widespread. The next section presents a short overview of the relationship between six major modifiable lifestyle factors and cognitive functioning.

Physical activity is associated with a lower risk of Alzheimer's disease or any type of dementia, and older people with better cardiovascular function, who are more physically active, have decreased chances of cognitive decline [11,12]. Already in 2007, a plea was made for physical activity trials as prevention for cognitive decline [13]. Furthermore, physical inactivity has been calculated to account for approximately 5.3 million premature preventable deaths in 2008, effectively decreasing global life expectancy by 0.7 years [14]. The Internet has proven to be a valid way of changing participants' physical activity levels [15]. Albeit in modest ways, average significant effect sizes of 0.14 [16], 0.16 [1,4], and 0.17 [1] can mean great benefits on a societal level.

Good physical fitness (positively) and higher body mass index (negatively) are related to academic performance as early as in third and fifth grade [17]. These effects seem to transfer to later life, with high blood pressure and central obesity being negatively related to global cognitive functioning in general and more specifically executive functions, visuomotor skills, and memory [18,19]. Although the exact mechanisms and functions that are affected still need to be established by future research, being overweight appears to provide additional risk for cognitive impairment. A recent review summarizes the positive effects of antioxidants and balanced nutrition on the delay and avoidance of onset of dementia [20].

Smoking is one of the most studied health behaviors, but only recently researchers have started to investigate whether smoking cessation has a positive effect on cognitive functioning. Even though results do not yet appear definitive, most research points towards current smoking as a risk factor for Alzheimer's and vascular dementia [21]. However, smoking cessation seems to mitigate the effects of smoking in the past, and relative risk of getting neurodegenerative diseases later in life decreases to normal levels [21,22]. Depending on the number of cigarettes a person smokes daily, the risk of various forms of dementia increases by 1.59 up to 2.72 times [23-25]. In addition to an increased risk of getting dementia, smokers generally have a lower level of cognitive functioning while smoking and experience faster decline as they age [26].

Alcohol consumption is not an unequivocal area in comparison with the behaviors discussed above. Low to moderate alcohol consumption may very well have positive effects on brain health, but too much alcohol is harmful to the brain. Cross-sectional studies show that moderate alcohol consumption (up to three units a day) may have beneficial effects on episodic memory, executive functioning, and processing speed of the brain [27-30]. However, these results should be interpreted with care. There

are no systematic or controlled-trial intervention studies available that examine the influence of alcohol consumption on cognitive functioning, but earlier research has shown that alcohol consumption higher than three units per day is harmful to the brain and can cause Korsakoff's syndrome [31]. In addition, it is not clear whether the positive effects on cognition are the direct result of the alcohol consumption itself. It may also be that people who have a lifestyle that includes moderate alcohol consumption also moderate themselves in other lifestyle areas, making them better cognitive agers. Further, a recent meta-analysis of epidemiological studies claimed that a reduction of 17% in alcohol consumption causes a 10% reduction in risk of cardiovascular diseases [32].

In a very elaborate review, Goel et al conclude that both acute and chronic sleep deprivation severely influence cognitive capabilities, starting with a measurable drop in performance on executive functioning tasks after being awake for 16 hours [33]. Among others, sleep deprivation further negatively influences psychomotor speed, learning and memory, and working memory performance, and causes faster performance deterioration on longer tasks [33]. Sleep deprivation by lifestyle choice, whether it is chronic or acute, affects executive functioning as the prefrontal and anterior cingulate cortices and posterior parietal systems are especially susceptible to sleep loss [33].

An increase in psychosocial stress can lead to burnout or depression, which negatively affects a person's cognitive functioning [34]. Among others, attention, concentration, flexibility, and memory deteriorate with higher amounts of perceived stress [19,35]. Epidemiological research shows a connection between the tendency to experience stress and the risk of mild cognitive impairment and Alzheimer's disease [36-38]. Also, the speed at which older people experience cognitive decline is correlated with the tendency to experience stress [36].

Managing these modifiable lifestyle factors could serve as a strong protective factor against neurodegenerative syndromes such as dementia. Stimulating health behavior change via the Internet appears feasible, and even the use itself of computers may serve as a protective factor when it comes to dementia [39]. Therefore, we plan to design an online, complex eHealth intervention aiming at lifestyle improvement with cognition as the primary outcome measurement. The research question for the current intervention with the Brain Aging Monitor (BAM) will be twofold: (1) Is the use of a self-motivated, complex eHealth intervention effective in changing multiple health behaviors related to cognitive aging in Dutch adults in the work force, especially those aged 40 and over? and (2) Does this health behavior change result in healthier cognitive aging patterns, thereby possibly preventing or delaying future onset of neurodegenerative syndromes like Alzheimer's disease?

Methods

The methodology of this study protocol follows the Standard Protocol Items: Recommendations for Interventional Trials (SPIRIT) guidelines that are specifically developed to provide guidance for researchers to document their study protocol and ensure that no relevant information is missing [40].

Study Design

For this study, we will use a quasi-experimental longitudinal pre-posttest design, with measurements at baseline and after 12 and 24 months. Only Dutch individuals within the Netherlands will be recruited. The intervention website is programmed in Dutch and as such will not be feasible for implementation outside the Netherlands. Since the Brain Aging Monitor is an eHealth intervention and the Netherlands has an Internet penetration of 94% of all households [41], there is no limit to its potential reach within the Dutch-speaking community. There are no regional restrictions that keep uptake of the intervention pinned to the region of the research institute. Since the entire intervention is based on self-management, contact with the research team is strictly limited to technical support. Considering the fact that the protocol relates to a pragmatic field study that will recruit both at an individual and organizational level, we cannot give an accurate estimate of the number of sites needed to obtain the necessary number of participants. For a sample size calculation, we refer to the description of sample size (Nederlands Trial Register: NTR4144).

Eligibility Criteria

The intervention will be performed among the general Dutch population and is aimed at delaying and/or slowing down cognitive aging. A recent study by Singh-Manoux et al showed that cognitive decline can be measured as early as 45 years of age [42]. For this reason, participants are eligible for analysis of the primary outcome if they are at least 40 years or older. The upper age limit is a more pragmatic one, since the intervention is aimed at the Dutch workforce and 67 is the official retirement age. Apart from this age criterion, a participant has to have sufficient comprehension of the Dutch language to understand the digital informed consent form (see Multimedia Appendix 1) and should have regular access to an Internet connection. Because the entire intervention takes place outside of the research facility, no strict control or enforcement is possible over other ineligibility criteria such as neurodegenerative disorders, medicine use, or psychiatric symptoms. Therefore, we decided not to use these and other possible exclusion criteria, which also increases overall external validity.

Health Behavior Change Theory

Using Lustria's organizing heuristic for strategies in computer-tailored online behavioral interventions, the BAM is an iterative, self-guided customized health program, with expert-led technical support [5]. The BAM uses different modalities such as Web content, email, online newsletters, and online games. To build self-regulatory skills, the BAM deploys goal-setting activities, skill-building activities, self-monitoring, and email reminders. According to a meta-analysis by Webb et al, applying a more extensive use of theory in online lifestyle tools increases overall effect size [4]. Theory can aid intervention designers identify appropriate targets for intervention, select intervention techniques, and it can illuminate which mechanisms of change are effective [43]. How theory is applied in the BAM will be described after the description of the intervention program itself.

Intervention

The BAM eHealth intervention website is open to anyone who is interested in the relationship between healthy living and brain aging. Figure 1 gives a short overview of the flow of a new participant within the intervention, and Figure 2 shows a screenshot of the BAM homepage. The BAM is a complex intervention [44], using multiple intervention components aimed at multiple health behaviors. As mentioned, the BAM focuses on physical activity, nutrition, smoking, alcohol, sleep, and stress. The BAM has an assessment and feedback system. After registering, validating their email address, and signing a digital informed consent form, new participants fill out seven short questionnaires (ranging from 4-20 questions or statements): one questionnaire for every lifestyle factor and one additional questionnaire on individual characteristics. Full lifestyle questionnaires and their references can be found in Multimedia Appendix 2. The answers to these questionnaires are used to create a personal lifestyle profile for the participant. The participant receives feedback per question using an easy-to-understand visual traffic light (green=conform to the norm, yellow=close to the norm, orange=much room for improvement, red=non-norm compliant) based on the health authority recommendations, behavior-specific feedback on health authority recommendations, and reference values on peer behavior (if possible and/or applicable divided for age and gender). Figure 3 shows a screenshot of the feedback on the nutrition questionnaire. This gives the participant a fast and detailed overview of their current lifestyle status. Also, the answers to the questionnaires tailor the intervention to the participant. For example, non-smokers will not be confronted with information about smoking or the option of setting goals that apply only to smokers.

The use of short self-reporting questionnaires on health behavior is a decision from a time-saving and retention perspective. Using more elaborate questionnaires would allow for better insight in a participant's behavior but will likely result in higher attrition [45]. Furthermore, more elaborate questionnaires are likely to pose questions that are difficult to answer for an individual. For example, obtaining a meaningful, valid answer on a participant's consumed dietary fiber (using the Dutch Healthy Diet index [46]) is very difficult. This would require a 24-hour recall process during the initial registering procedure, risking immediate dropout. Therefore, we chose to use simple questionnaires for all lifestyle areas, where every question covers a behavioral trait that directly relates to a goal that can be set later on in the program.

After the questionnaires, the Brain Aging Monitor Cognitive Assessment Battery (BAM-COG) opens up on the game wall and the Goal-Setting Module (GSM) is unlocked. The BAM-COG is an online cognitive assessment battery that has been specifically developed for use in the BAM and has been validated by our group [47]. These games measure working memory, visuospatial short-term memory, episodic recognition memory, and planning. An instructional arrow will direct the participants' attention to the fact that the games are open for play. After receiving their personal lifestyle overview, participants can start setting monthly, personal-health behavior goals using the GSM. We based the GSM on the Goal Attainment Scaling (GAS) methodology by Kiresuk [48]. Using the GAS triggers participants to be more conscious about their goals because it does not rely on a single digit. Instead, it requires the participant to fill out a complete scale from -2 to 2 (where -2="I have made minimal progress", 0="I have reached my original goal", and 2="I have done a lot better than my original goal"). This not only requires more attention from the participant while setting the goal, but it also enables the BAM to give positive feedback on partially accomplished goals instead of a "yes" or "no" answer to the question "did you reach your goal?".

Every potential goal is accompanied with a set of instructions guiding the participant towards personally relevant and realistic goal setting. It starts with an example GAS scale, a case of a fictive participant, and a step-by-step instruction to complete the goal-setting process. The GAS system is programmed to return a number of restrictions or error messages to the user: (1) if values overlap, (2) if values are in the wrong direction or scrambled, (3) if values exceed the value of 7 days per week, or (4) if the given 0-value is a step back from the value that was answered during the intake questionnaire. When the goal is set, the participant is given reinforcing feedback on making a good first step towards behavior change. After this message, a list of instructions and tips are given that are relevant for that specific goal. This list is open for the participant to choose their preferential working method and go from very basic instructions (eg, "buy fruit" in case of a goal "eat more fruit") to signing social contracts or using implementation intentions (eg, "if there is no running group in the neighborhood, then I will start my own running group" in case of a goal "start to work out") [49].

After a participant has decided which instructions and tips to use, the goal gets transferred to the short-term monitoring system (STMS). Here, participants can monitor their own behavior on a day-to-day basis. Inputting their behavior in the STMS, the system automatically graphs a quick overview of how well a participant is doing for that goal on a week-to-week basis. After a month, the STMS asks the participant to what extent the goal is accomplished on their own original GAS scale. For any score specific to that goal, the BAM gives tailored feedback. If a score of -2 or -1 is obtained, the goal is deleted from the participant's profile and encouragement to try again is given. However, if a score of 0, 1, or 2 is obtained, the goal gets transferred to the long-term monitoring system (LTMS). In the LTMS, a participant gets monthly follow-up questions to monitor if they still are maintaining their initial level of behavior change. With every monthly question that gets answered, the participant is given tailored feedback to acknowledge their success or motivate the participant to maintain their initial behavior change. Multiple goals can be graphed over time giving a personal overview of all acquired and maintained behavior change goals. If multiple goals are set on the same subject, new goals will overwrite old goals so that only the most up-to-date information is shown. Because the BAM does not dictate how many a goals a participant sets and in what order they do this, it implicitly provides the participant with a conceptual choice between simultaneous or sequential goal setting, giving every participant the option to work at their own pace and preference [50-52].

In order not to overload the participant with questionnaires immediately after registering, the personality questionnaires become available to the participant 7 days after registration. These are the Dutch General Self-Efficacy Scale [53], lifestyle factor specific self-efficacy questions [54], the Positive Affect Negative Affect Scale [55], and the Self-Control Scale [56]. These questionnaires are administered to perform secondary analyses to check if the BAM is more effective in certain personality types. No feedback on the personality questionnaires is provided for the participants.

After all baseline data are collected, the tailored knowledge databases, the buddy system, healthy recipes, and blogs on health behavior and cognitive aging become available. The buddy system is a built-in control mechanism the BAM uses to ensure goal safety. A subset of goals that are suitable for this purpose are anonymously sent to another BAM user to be judged by a BAM buddy on its feasibility and safety. A buddy can judge a goal to be “ambitious”, “not ambitious enough”, “just right”, or in the case of losing weight, “this seems unhealthy”. This gives the goal-setter an opportunity to get instant feedback on the feasibility of their goal. Also, it gives the buddy a “look behind the scenes” that may provide feedback on their own goal-setting behavior, as the exact same situation for somebody else may be perceived as harmful whereas this same goal would be deemed applicable to the participant themselves. At the same time that the buddy system becomes available, participants also get access to the knowledge databases that contain up-to-date information on healthy living and the relationship between the different behaviors and brain health. Last, participants get access to weekly blogs on lifestyle, research and brain aging, and healthy recipes. After 365 and 730 days upon completion of the baseline measurement, they will be recruited for 1-year and 2-year follow-up measurements.

No reasons for discontinuation of the study of a participant by the research team have been specified. There is no disease load that can be exacerbated by participating in the BAM nor is there any medication prescribed that could have negative health consequences. Participants are provided with the option of unsubscribing to the study at any given time in their personal profile space. When a participant decides to leave the program, a short questionnaire is automatically presented to collect data on the reason for unsubscribing and to inquire if the participant may be approached to partake in the 1-year and 2-year measurement, regardless of subscription status. All of this is voluntary and participants can always choose to skip this questionnaire.

Keeping participants engaged with eHealth intervention programs has been a major problem since the field originated [7,57,58]. Several adherence-enhancing strategies are in place with the BAM. First, we upload weekly news updates on the homepage regarding health behavior and brain aging from the largest Dutch news websites. Second, on the dashboard of the participant we will upload weekly blogs and healthy recipes so that the content, apart from user input, changes on a weekly basis. Blogs discuss current topics in research on anything BAM-related in an easy accessible form. Recipes use fresh and healthy products and provide participants with ideas to prepare a healthy meal. Third, a personalized email reminder system is built into the BAM that can be adjusted to the participants’ individual needs. In their personal profile space, a participant can choose to receive daily, weekly, biweekly, or monthly email reminders. These reminders give an overview of current active goals and will link the participant directly to this goal after logging in. Fourth, during the registration process new participants can opt in to receive BAM newsletters, which will be sent using the MailChimp engine. At any time, participants can opt in or opt out of the newsletter. Last, participants get a personal profile space with a quick and easy overview of their current lifestyle. They can make adaptations to their lifestyle, see the results of their goals, and can adjust their settings.

Considering the field setting for this intervention, it is difficult to control for concomitant care, or better yet if the BAM inspires participants to make use of other platforms to alter their health behavior in a positive way, this accomplishes the BAM’s goals. The BAM can and may function as a gateway to healthy behavior. Using the GSM and the yearly follow-up, the BAM can track changes over time even if participants actively use outside help that the BAM refers them to.

Figure 1. Flowchart of process a new participant goes through upon registration.

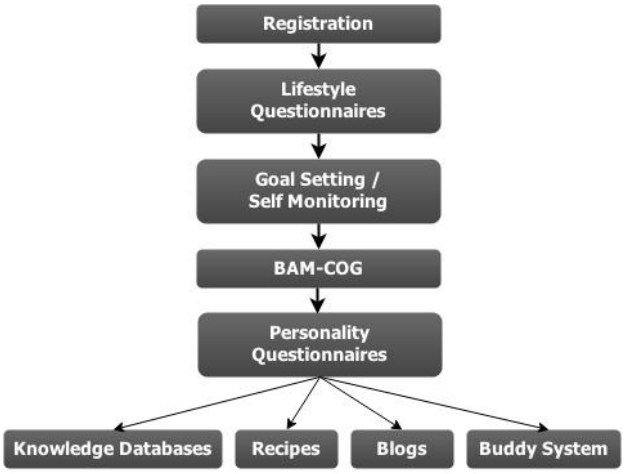


Figure 2. Brain Aging Monitor homepage.

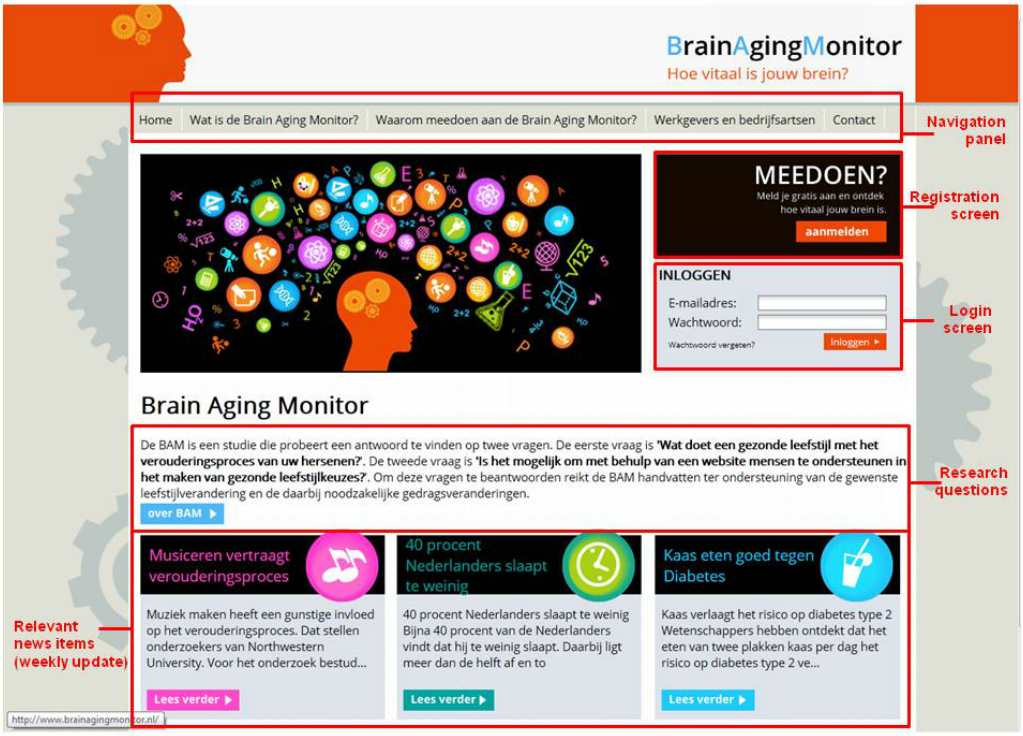
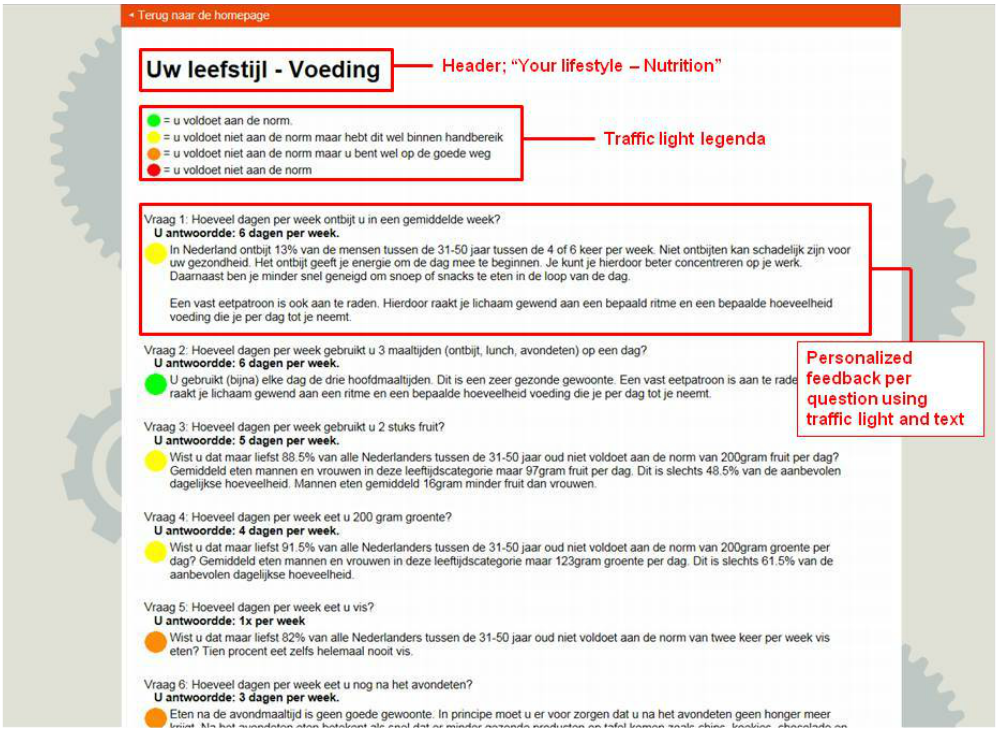


Figure 3. Screenshot of feedback module on nutrition questionnaire.



Use of Health Behavior Change Theory

A description of the behavior change techniques used is given using the taxonomy provided by Abraham and Michie [59]. This paper provides researchers with 26 behavior change techniques based on behavior change theories to be used while designing an intervention. As the BAM is a self-guided, voluntary online intervention, the majority of participants will be in the last three stages of the Transtheoretical Model (TTM): the preparation, action, and maintenance phases [60]. The BAM guides participants from preparation phase (informing) to action phase (goal setting, short-term self-monitoring) to maintenance phase (long-term self-monitoring). As participants enroll in the BAM, the three stages of the TTM are facilitated instantaneously. And for every behavior, the participant can choose the most appropriate phase to start with. When the preparation phase is chosen, the knowledge database for that

behavior is the most suitable starting point. Providing a participant with information is in accordance with the Information-Motivation-Behavioral Skills Model (Fisher).

When participants decide to enter the action phase of the TTM and become active goal-setters, they get an appropriate list of instructions and tips. Setting active behavioral goals is a part of most renowned behavior change theories such as the Theory of Planned Behavior (TPB) [61] and the Social Cognitive Theory (SCT) [62]. In line with the SCT, general encouragement is provided when goals are set and (partially) accomplished. While working with the tips and tools, participants will monitor their behavior. Bandura notes in his SCT that self-directed change can be promoted with features that allow program participants to set realistic goals, provide them with instructions and tips to reach these goals, and to allow for detailed self-monitoring of their own behavior over time [63,64]. Using GAS prompts a participant to set specific lifestyle goals, the buddy system prompts the review of behavioral goals, the short-term monitoring system prompts self-monitoring of behavior, and evaluating their own performance gives the participant feedback on their own behavior. The instructions and tips, when of additional value, include a list of commonly heard behavioral barriers and reasons why these barriers are either invalid or on how to overcome them, which is part of the SCT. Another possible instruction was to set up a behavioral contract with a person close to the goal-setter to create a form of peer pressure or control, which is in line with the theory of operant conditioning [59]. Among the instructions and tips, when applicable, is the suggestion to form “if-then” implementation intentions [63]. These if-then implementation intentions partially ease the transformation of behavior but also aid in relapse prevention, as these actively trigger the goal-setter to identify risk situations and come up with an appropriate action if the occasion arises (eg, IF my friends keep asking me to drink beer, THEN I will firmly tell them that I am drinking water this evening). Finally, the goal that can be set to reduce stress and optimize satisfaction with life aims at stress reduction through various methods (eg, yoga, mindfulness).

After the completion of a goal, follow-up is built into the system by automatically re-evaluating changed behavior on a monthly basis. Also, using an automated email reminder system, the BAM aims to maximize adherence to the program, providing follow-up prompts. To summarize, in accordance with the taxonomy by Abraham and Michie, 13 out of 26 behavior change techniques are used in the BAM (#1, 4, 5, 6, 8, 10-13, 16, 18, 23, and 24) [59].

Research Questions and Outcome Measures

Our primary research questions are the following: (1) Is successful health behavior change related to better cognitive aging patterns over time? Change scores will be calculated by subtracting baseline scores from scores at year 1 and year 2, (2) Does the BAM facilitate health behavior change? (see Multimedia Appendix 3 for the construction of the overall lifestyle score), and (3) Does the BAM facilitate health behavior change in certain specific lifestyle areas better than others?

Our secondary research questions are as follows: (1) Is there a dose-response relationship between the number of goals participants set and the expected amount of health behavior improvement? and (2) Does the BAM increase feelings of self-efficacy in health-related behavior and a change in self-control scores from baseline to 1 and 2 years of intervention, as measured with the Self-Control Scale [56]. In other words, does the BAM increase feelings of being in control of one’s life?

Our primary outcome measures are (1) cognitive change over time, (2) overall lifestyle change over time, and (3) specific lifestyle changes over time. The secondary outcome measures consist of (1) number of goals set, (2) change in self-efficacy, and (3) change in self-control.

Participant Timeline

The timeline for the BAM is straightforward (see Figure 1). Participants need to register only once. Immediately after registering, email validation, and the informed consent form, the lifestyle questionnaires are available. Once the lifestyle questionnaires have been completed, the BAM-COG becomes available. Seven days after subscription, the personality questionnaires appear in the personal dashboard. These features are all presented again to the participant 365 and 730 days after their baseline completion. The GSM, STMS, and LTMS are continuous processes from the moment they first become available to the participant. After 1 year (365 days), the data will be collected for preliminary secondary outcome analysis. After the 2-year follow-up (730 days), the data will be used for analysis of both primary and secondary outcomes. The nature of the grant requires that the intervention remain online even after data collection is finished for the initial study period and that the BAM remain open to the public after the study is completed. Adaptations to the program can be made at this time, according to study outcomes.

Sample Size

We aim for a group size of 200 to find a 15% reduction on the risk factors (power calculation based on alpha <.05; power of 0.8; two-tailed; n=166; ±20% dropout).

Recruitment

Different recruitment strategies will be implemented to reach the necessary sample size. First, we will recruit medium to large commercial or governmental organizations through their human resources department or company employed medical staff. The BAM can provide organizations with a concrete intervention program that can substantiate their health policy. Organizations will be recruited by direct inquiry through telephone calls, emails, and will be targeted during several symposia, workshops, and congresses where the BAM will be presented. Once an organization is recruited, the research team in collaboration with the human resources department will develop a tailored recruitment strategy that maximizes the use of existing communication channels within the organization. These organizations are expected to deliver approximately 50% of all study participants.

Next to organizational recruitment, the BAM will also recruit participants in the general Dutch population. A press release will be issued by the Radboudumc to reach mainstream media

to generate national attention for the study. The BAM will be advertised at the website of a cooperative research consortium that draws national attention because researchers from four nationally spread out universities will promote this website. Also, we will present the BAM at national and international health care conferences. We estimate that approximately 50% of all study participants will result from this free recruitment strategy.

The enrollment period will take approximately 4-5 months. However, due to the nature of the grant no actual stop in participant influx will be enforced. Participants will be allowed to enter the study at any given time. We will monitor participant influx over time so we can keep estimating the relevance and need for an intervention such as the BAM. Unless individual organizations determine otherwise, no financial incentives will be offered to potential participants. If this occurs, this will be disclosed in future publications.

Sequence Generation, Allocation Concealment Mechanism, Implementation, and Blinding

This pragmatic field trial does not use a control group. Therefore, sequence generation, allocation concealment, implementation, and blinding are not discussed.

Data Collection Methods

Data collection in the BAM is completely automated through its website. Therefore training of personnel is irrelevant. All data collection forms will be equal for each new participant who subscribes to the program. For collection of the descriptive lifestyle data, questionnaires have been used that accurately represent the relevant health norm or health behavior (full lifestyle questionnaires can be found in Multimedia Appendix 2). For measuring cognitive functioning, the BAM program uses a validated online self-monitor for cognitive functioning, specifically developed for use in the BAM, called the BAM-COG [47]. We have deliberately chosen to keep the baseline assessment as concise as possible. Creating a complete overview of a participant's lifestyle can be a tedious task and with high risk of early dropout in eHealth interventions, the BAM's lifestyle assessment is meant to give a fast and easy overview of a participant's compliance to health norms, not a detailed description of all facets that make up healthy living.

The BAM intervention has multiple built-in mechanisms aimed at increasing retention to protocol. As described in the intervention section of this protocol, we will use blogs and recipes in the intervention to keep participants' focus on the program, as well as the deployment of the adjustable reminder system and the flexible use of newsletters. When the program has been online for 365 and 730 days, special newsletters will be sent out to all active participants reminding them of their annual follow-up measurement that becomes available on their personal dashboard.

Participants who want to exit the study can do so at any time. They can unsubscribe from their personal profile page using the unsubscribe instructions. Once this process is initiated by the participant, a short questionnaire will be used to identify the reason for dropout. Also, the BAM will ask the participant if they are still willing to be reminded of the annual measurements. In this case, they would not actively participate in goal setting and behavior monitoring but would be willing to come back and provide the program with follow-up data.

Data Management

Because of the eHealth nature of the intervention, all data entry and collection are done online and therefore are programmed to be completely automated. The intervention website is secured with up-to-date online security protocols and certificates safeguarding private information of participants. Users must sign in to get access to their profile and logged data. To sign in, a user name (email address) and password are required. Passwords are stored by using the MD5 hash algorithm. Each user gets their own session after signing in. This session will be killed when the user closes the browser or when the session times out. All data are stored in a MySQL database. To access this database, a password is required that contains digits as well as characters, randomly created. The site uses the HTTPS protocol and is secured by a Comodo SSL Certificate.

Data storage will be extensively tested in the pilot phase. All the participants are assigned an anonymous personal identifier that will be used for all the tables containing data during data collection in the MySQL database. The data will be stored on secure hosting servers for 20 years after the completion of the intervention period.

Statistical Analyses

Intervention Effects

Primary analyses will be unadjusted. Depending on the distribution of continuous, categorical, and interval outcomes, an appropriate distribution and relevant statistical models will be used. These models will assess intervention effects at end-of-intervention (1-year and 2-year) as well as the difference between 1- and 2-year measurements. Baseline characteristics will be compared between groups using *t* tests for continuous variables and chi-square tests for categorical variables. Mann-Whitney tests will be used when baseline data are not normally distributed. If necessary, multivariate analysis, including multi-analysis of variance and multiple regression, will be performed to adjust potential confounders, including baseline demographic and behavioral characteristics. The covariates associated with outcomes or contributing to a significant part of variation of used multivariate models will be adjusted as potential confounders. The final model will include these covariates or remove those that do not affect estimates, if models show evidence of overfitting.

Secondary Analyses

Total set goals and specific lifestyle area goals will be reported descriptively. The changes of lifestyle within the goal setting group will be reported in absolute difference at end of intervention. The dose-response association between goals and change of lifestyle will be analyzed by multivariable linear regression model, and potential confounders will be adjusted. The change in self-efficacy and self-control scores will be reported in absolute difference at the end of the intervention. The association between the use of BAM and increased feelings

of self-efficacy or self-control will be analyzed by multivariate analysis.

Analysis will be performed per protocol. The dropout in this pragmatic field study is likely to be high but can be considered a separate outcome for the implementation and feasibility of these types of intervention. As such, it represents valuable data about the quasi-experimental study design.

Data Monitoring

There will be no data monitoring committee for this intervention, as no adverse events are expected. No interim analysis will be performed for the same reason. No significant harm to the participants is to be expected for the BAM intervention program. If anything would occur, participants can contact the research team through the contact form on the website. Standard data monitoring procedures for the scientific Geriatric Medicine department at the Radboudumc apply.

Auditing

No auditing is planned specifically aimed at the BAM study. However, the BAM is part of the scientific branch of the Geriatric Department of the Radboudumc and therefore can be routinely audited internally.

Research Ethics Approval and Protocol Amendments

The program is largely implemented in medium-to-large corporations that transparently implement the program as part of their health policy. From individual participants, an online informed consent form for their participation in scientific analysis is obtained during registration for the program. This study was deemed exempt from formal ethical evaluation by the local medical ethics committee (region Arnhem-Nijmegen, registration number: 2014-1268). Protocol amendments will be submitted, if necessary.

Informed Consent

Due to the online nature of this study, no personal informed consent can be obtained from participants. However, we do use an online informed consent form to make sure that participants are aware of their participation in scientific research. Therefore an extra step has been added to the registration process. After email verification, before a participant can start the program, a screen appears with an informed consent form. If informed consent is not provided by ticking the correct box, participation in the program cannot continue. See Multimedia Appendix 1 for a complete translation of the information provided and accompanying informed consent form.

Confidentiality

All information stored in online databases is random-password protected. Also, all our websites use state-of-the-art SSL-security certificates to ensure maximum safety of participants' confidential information. MySQL data that contain names are stored in different tables as study results. Exported data from the MySQL online databases will be downloaded to local password-protected hard drives for analysis. Also, when databases are saved on local hard drives, these databases will be stored anonymously, using only anonymous personal identifier codes for all participants. No print records will be kept at any point during the study. Participants' study information will not be released outside of the study without the documented permission of the participant.

Access to Data

Only principal investigators, post-docs, and PhD students involved in the study will have access to the full raw dataset. Other researchers interested in using the BAM dataset will get access to a cleaned dataset. Human resources departments of recruited companies will, at no point, get access to any form of dataset. They will receive anonymous overall group results of data analysis.

Ancillary and Post Trial Care

Participants can always contact the research team by phone or email with any questions they may have. Also, the BAM will remain available to them after the study closes since the BAM is part of a national Quick Results grant aimed at providing fully functional end products at the end of the study period.

Dissemination Policy

Study results, regardless of their direction of outcome, will be published in high standard, peer-reviewed scientific journals. Study publications will be written on primary and secondary outcomes and subgroup analysis. Researchers and health care professionals will be updated on study results during national and international conferences and workshops and with targeted tailored publications in relevant professional magazines.

Participants who unsubscribe to the program are given the option to stay updated on study results when these become publicly available in the form of a summary of the research report or PhD thesis. Participants who stay in the program until the study period closes will be approached by email to probe their interest in study results. Furthermore, study results will be made available to the general public via a press release issued by the Radboudumc after publication of the primary outcomes. No publication restrictions apply, and no ghost- or professional medical writers are involved in the study.

Results

Study results are expected to be published early in 2016.

Discussion

Principal Considerations

To our knowledge, this is the first large study that aims at health behavior gains with a cognitive motivation and outcome measure in the general population. Furthermore, the BAM is one of the first studies launched in an era when almost everybody has an Internet connection. As such it can serve as a proxy for the feasibility of these types of interventions when specifically launched in the general population.

Enhancing the BAM with state-of-the-art, scientifically validated applied games gives the unique advantage of being capable of measuring cognitive functioning while maintaining all the advantages such as reach and low cost that are associated with eHealth studies. The use of online applied games from the safety

and comfort of one's home gives us another major advantage. It also provides a motivational edge, since playing games is considered more appealing than participating in standard neuropsychological testing. We decided to use self-reporting measures for the BAM instead of more objective clinical measures (eg, blood pressure, cholesterol) because use of these measures in a general community dwelling research population is either not feasible or expensive and a big logistic challenge. Moreover, using a participant's self-reported input closely matches the participant's perception of their own behavior. Therefore, the goals a participant sets are more likely to be perceived as personally relevant and the participant will feel ownership over the goal and behavior change that needs to be achieved. Combined, we feel this increases the odds of successful implementation of the BAM.

We chose a quasi-experimental design as it seems more appropriate for a field setting in which it is highly impractical to initiate a randomized controlled trial, and blinding participants to the type of eHealth intervention they are receiving is practically impossible [65]. Theoretically, it was preferable to use a step-wedge cluster-randomized controlled design, but this was not feasible with the current 2-year intervention period. Since recruitment of companies is not guaranteed, cluster randomizing from the start is also difficult, especially since organizations are not very likely to see the incentive of participating as a control group. There is also a pragmatic side to the choice of the population. There is substantial theoretical background to select participants aged 40 and older [42], since in this part of the working population cognitive decline can already be measured, and they are more likely to be triggered by a dementia prevention program. People under 40 are less likely to be triggered by cognitive decline or even dementia prevention, as it is a disease associated with old age and only in later years a relevant threat to their health. Nonetheless, the BAM will allow participants under 40 to subscribe. However, lifestyle advice will be tailored to age cohorts starting at age 40.

Last, the use of a multimodal lifestyle perspective is a strength, as it gives potential participants a more integral overview of lifestyle. Providing a more comprehensive lifestyle overview allows the participant to prioritize one type of change over another and take a holistic approach to their own lifestyle. Also, benefits from changing one behavior may transfer to improved outcomes on another behavior that would go unnoticed in single modal interventions. Since the BAM is an eHealth intervention, tailoring to the needs of the participant is cheap after initial development costs have been incurred. Targeting personally relevant lifestyle factors after providing a more general overview should improve program adherence because the participant becomes aware of why they are working on a certain risk factor. This is important as adherence is often the crucial factor in lifestyle improvement programs.

Conclusion

This study will add to the body of evidence on the effectiveness of eHealth intervention programs with the combined use of state-of-the-art applied games and established behavior change techniques. This will lead to new insights on how to use behavior change techniques and theory in multidimensional lifestyle eHealth research, and how these techniques and theories apply when they are used in a setting where no professional back-end is available.

Acknowledgments

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Authors' Contributions

MOR conceived of the study and participated in the design of the study. TA and MAEB initiated the study design and drafted the manuscript. LQ, AdL, and RCPK aided in designing the study and revised the manuscript. All authors contributed to refinement of the study protocol and approved the final manuscript.

Conflicts of Interest

None declared.

Multimedia Appendix 1

Translated version of the informed consent form.

[PDF File (Adobe PDF File), 15KB - resprot_v4i4e130_app1.pdf]

Multimedia Appendix 2

Questionnaires.

[PDF File (Adobe PDF File), 24KB - resprot_v4i4e130_app2.pdf]

Multimedia Appendix 3

Construction of the overall lifestyle score.

[PDF File (Adobe PDF File), 4KB - resprot_v4i4e130_app3.pdf]

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Abbreviations

BAM: Brain Aging Monitor
BAM-COG: Brain Aging Monitor – Cognitive Assessment Battery
GAS: Goal Attainment Scaling
GSM: Goal-Setting Module
LTMS: Long-Term Monitoring System
SCT: Social Cognitive Theory
SPIRIT: Standard Protocol Items: Recommendations for Interventional Trials
STMS: Short-Term Monitoring System
TPB: Theory of Planned Behavior
TTM: Transtheoretical Model

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Original Paper

An Internet-Based Intervention (Mamma Mia) for Postpartum Depression: Mapping the Development from Theory to Practice

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Abstract

Background: As much as 10-15% of new mothers experience depression postpartum. An Internet-based intervention (Mamma Mia) was developed with the primary aims of preventing depressive symptoms and enhancing subjective well-being among pregnant and postpartum women. A secondary aim of Mamma Mia was to ease the transition of becoming a mother by providing knowledge, techniques, and support during pregnancy and after birth.

Objective: The aim of the paper is to provide a systematic and comprehensive description of the intervention rationale and the development of Mamma Mia.

Methods: For this purpose, we used the intervention mapping (IM) protocol as descriptive tool, which consists of the following 6 steps: (1) a needs assessment, (2) definition of change objectives, (3) selection of theoretical methods and practical strategies, (4) development of program components, (5) planning adoption and implementation, and (6) planning evaluation.

Results: Mamma Mia is a fully automated Internet intervention available for computers, tablets, and smartphones, intended for individual use by the mother. It starts in gestational week 18-24 and lasts up to when the baby becomes 6 months old. This intervention applies a tunneled design to guide the woman through the program in a step-by-step fashion in accordance with the psychological preparations of becoming a mother. The intervention is delivered by email and interactive websites, combining text, pictures, prerecorded audio files, and user input. It targets risk and protective factors for postpartum depression such as prepartum and postpartum attachment, couple satisfaction, social support, and subjective well-being, as identified in the needs assessment. The plan is to implement Mamma Mia directly to users and as part of ordinary services at well-baby clinics, and to evaluate the effectiveness of Mamma Mia in a randomized controlled trial and assess users' experiences with the program.

Conclusions: The IM of Mamma Mia has made clear the rationale for the intervention, and linked theories and empirical evidence to the contents and materials of the program. This meets the recent calls for intervention descriptions and may inform future studies, development of interventions, and systematic reviews.

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KEYWORDS

early intervention; Internet; intervention mapping; Mamma Mia; postpartum depression; pregnancy; well-being

Introduction

Background

The postpartum period represents a vulnerable time where the woman is at increased risk of mental disorders [1]. Between 10% and 15% of women experience moderate to severe depressive symptoms during pregnancy and after childbirth [2-5]. Depressive symptoms postpartum can have severe consequences and lead to negative parenting behaviors [6], child psychopathology in general [7], and increase the risk of depression among partners [8]. The prevention and treatment of postpartum depression (PPD) is thus essential for a mother, her infant, and the family's mental health and well-being.

Psychological treatments for PPD are effective [9]; however, many women living with PPD are not identified and do not receive adequate treatment (see eg, [10]). This is a serious concern not only for the affected families, but also for the society as a whole. The social costs of mental illnesses for each annual cohort of births are estimated to be approximately £1.2 billion [11]. Consequently, there is a need to reach pregnant women and provide accessible evidence-based help and support to prevent PPD.

Internet interventions may be feasible in preventing and treating PPD with its potential for high reach. In fact, many pregnant women use the Internet to search for pregnancy-related information such as fetal development or childbirth [12]. Trend data show that 66-75% of Norwegian women in childbearing age (ie, 16-44 years) searched for health information online in the past 3 months in 2013 [13], and in the United States, 19% of Internet users report to have searched for information about pregnancy and childbirth [14]. Many women with PPD also express an interest in Internet interventions and report that they would use the Internet to learn coping strategies for PPD [15].

Recent studies have demonstrated the acceptability and feasibility of Internet interventions for PPD [16-20]. Results from the first randomized trials also offer promise for Internet interventions as an effective treatment for PPD [21-23]. However, as in almost all intervention research, intervention descriptions tend to be rather brief and general, and often confined to a few paragraphs in the methods section. This makes it difficult for researchers to identify active ingredients and practically impossible for intervention designers to make informed decisions about future intervention development and how to improve existing interventions. This is a serious concern for intervention research because it violates one of the basic premises of research—*replication* of studies. In other words, the reporting of interventions and conduct of intervention studies generally fail to contribute toward a cumulative science of Internet interventions.

Objective

The aim of this paper was therefore to provide a systematic and comprehensive review of an Internet intervention for the prevention of postpartum depressive symptoms and enhancement of subjective well-being. While previous research has mainly focused on the treatment of PPD, this intervention has a strong preventive focus considering the potential for social savings (see the earlier discussion). We used the intervention mapping (IM) protocol, which outlines the path from recognition of a need or problem to the identification of a solution [24]. The end product constitutes a comprehensive blueprint of the intervention and a detailed treatment rationale that may facilitate replication, support the interpretation of subsequent implementation and evaluation studies, and ease the comparison of the treatment rationale across interventions [25,26].

Methods

Intervention Mapping

IM is a tool that systematizes and integrates theory, empirical evidence, and information collected from the target population when designing health promotion programs. It makes the development of interventions transparent and provides an explicit report of all the decisions and considerations throughout the intervention process. There are 6 fundamental steps in the IM process: (1) the conduction of a needs assessment or problem analysis; (2) the definition of proximal program objectives based on scientific analyses of a given health problem and its predictors; (3) the selection of theory-based intervention methods and practical strategies to change (determinants of) health-related behaviors; (4) the production of program components, design, and production; (5) the anticipation of program adoption, implementation, and sustainability; and (6) the anticipation of process and effect evaluation. Each step comprises several tasks and the completion of 1 task guides the completion of a subsequent task. Although IM is presented as a series of steps, Bartholomew and colleagues [24] look at the planning process as iterative rather than linear, meaning that program planners move back and forth between the various tasks and steps. The process is also cumulative in the sense that each step is based on previous steps, and the failure to attend to important aspects in any given step may lead to mistakes and inadequate decisions in subsequent steps.

Post Hoc Application of the IM Protocol

The IM framework greatly influenced the development of Mamma Mia, which makes a post hoc analysis both feasible and informative. Ideally, the IM protocol is used a priori in intervention development. In this study, however, the IM procedure was applied in a post hoc manner. Previous studies illustrate, though, that a retrospective IM-based analysis can also be a useful tool for post hoc description of interventions [27,28]. Specifically, it may point toward weaknesses in the intervention development process and the intervention itself, thereby anticipating any potential threats and issues that may arise during the implementation and evaluation. Nonetheless, an application of the IM protocol after the development of the intervention has taken place means that the actual course of actions deviate to a certain extent from what is prescribed by the IM protocol. Most notably, this concerns Steps 5 and 6 in the IM protocol where program adopters and implementers were not included in the intervention development in the strictest sense of the IM protocol; additionally, the evaluation of the intervention is mostly focused on the effectiveness of the intervention, rather than process evaluations of the development.

Any deviations from the IM protocol are noted throughout in the results.

Results

Step 1: Needs Assessment

A thorough exploration of the health problem, referred to as the needs assessment, is an inherent part of the IM framework. The result of a needs assessment illustrates how prevalent the problem is and what factors are associated with it. In this study, the health problem is PPD, and the challenge is that many women who experience depressive symptoms receive no counseling or support. An exploration of the literature suggests that many women report to be unfamiliar with symptoms of PPD and do not realize that they may be suffering from depression [29]. Symptoms of PPD may be difficult to distinguish from symptoms normally observed in postpartum women such as tiredness, changes in sleep, appetite, and sexual desire (ie, symptoms that are normally observed in women after giving childbirth and taking care of a newborn baby), making it also difficult for health professionals to detect women with PPD. This may explain, in part, why women often fail to seek help for their PPD (ie, 17-25%) [30]. Other barriers to help seeking include women's inability to disclose their feelings, for example, because of shame or fear of losing custody, and health professionals' reluctance to respond to the mothers' emotional and practical needs [31]. In addition, consultations with general physicians (GPs), midwives, and public health nurses (PHNs) at well-baby clinics tend to be rather brief in the prenatal and postnatal periods (ie, regular appointments in Norway are scheduled to last about 15-20 minutes), thereby making it difficult to detect and respond to PPD. A preventive intervention, however, can not only prevent the development of depressive symptoms, but also help a woman become aware of and identify symptoms of depression, and possibly encourage her to seek help and support.

As in any preventive intervention, it is important to target risk and protective factors that may influence the onset and development of PPD. However, most studies have typically emphasized and identified risk factors that are hard or even impossible to modify such as a previous history of depression, negative life events, and certain demographic characteristics [32]. Thus, as part of the current needs assessment, we conducted 2 studies to investigate the contribution of modifiable psychological risk factors associated with perinatal depressive symptoms and well-being further. In a longitudinal study, self-efficacy, certain cognitive emotion regulation strategies (eg, rumination, self-blame, and positive reinterpretation), perceived available support, and need for support were found to predict the rate of postpartum depressive symptoms [33]. Interviews with new mothers largely confirmed these findings, but also highlighted that the woman's expectations and approach to motherhood influenced her feelings of depressed mood and well-being. Specific expectations and a high need for mastery and planning (ie, controlling) made women more vulnerable and at-risk for experiencing lower mood and subjective well-being, compared with women who were more relaxed [34]. From an intervention perspective, it thus became important to target these modifiable psychological risk factors.

A recent and large Norwegian population-based study showed that relationship satisfaction protects against emotional distress during pregnancy [35], whereas dissatisfaction with the partner relationship predicted maternal emotional distress [36]. A satisfying relationship is important to prevent depression and to retain and increase life satisfaction [37], especially because both relationship and life satisfaction tend to decrease after childbirth and remain below prebirth level for several years [38]. Although Norwegian women may be more satisfied with their lives during pregnancy and following birth in general than women in other countries, it is still common that life satisfaction drops after the "baby honeymoon" period [39]. Hence, improving relationship satisfaction and well-being are major targets for preventing PPD in both pregnant and postpartum women.

Finally, parental insensitivity, which refers to disengagement, intrusiveness, or noncontingent responding, to the infant's cues is associated with PPD [40,41]. Increasing parental sensitivity thus makes up a final key modifiable psychological factor that should be targeted in an intervention. This is important because lack of parental sensitivity and insecure attachment relationships can instigate cycles of transactional or bidirectional effects that can both exacerbate parental symptoms of depression and increase the risk of internalizing and externalizing problems in infants [42-44], difficulties that may continue into late adolescence [45,46]. Prenatal depression and PPD may affect parenting capabilities such as parental sensitivity, which may in turn instigate the development of an insecure mother-child attachment relationship [47,48]. Thus, promoting healthy and supportive attachment relationships between parents and their infants, thereby increasing parental sensitivity, may be of great importance for parents and infants' long-term adjustment and mental health.

Step 2: The Performance and Change Objectives of Mamma Mia

The second step in IM is about defining the overall goals of an intervention and the performance and change objectives, which, in turn, specify how the overall goals can be achieved. To arrive at these objectives, the IM protocol suggests a procedure in which the overall goals are broken down into subgoals (ie, performance objectives) and correlates of subgoals are identified (ie, determinants). Change objectives are then constructed to target the determinants of the performance objectives. In short, change objectives are essentially what the user has to change or learn to attain the performance objectives.

The Overall Goals of the Intervention

The intervention (Mamma Mia) was designed as a universal preventive measure that could be offered to all pregnant women with the primary goals to (1) prevent the onset or development of depression and (2) enhance subjective well-being during the prenatal and postnatal period (ie, starts in gestational week 18-24 and lasts up to 6 months after giving birth). A secondary goal was to ease the transition of becoming a mother by providing knowledge, techniques, and support during pregnancy and after

birth. The reason for starting in the second trimester is that expectant mothers can then be reached as early as possible, they have a reduced risk of spontaneous miscarriage, all women attend ultrasound at this time (ie, see their baby) and start forming prenatal attachment to their baby, and because the prevalence of prenatal depression is as common as postpartum. Six months after birth is the end point of the intervention as postpartum depressive symptoms tend to fade around this time [1]. However, to prevent postpartum depressive symptoms and increase well-being are not complete descriptions of the desired outcome. The specific behaviors required to accomplish the desired outcomes need to be described in greater detail. To do so, we need to consider some basic facts about the psychological process of expecting a child and taking care of a newborn baby.

The Performance Objectives

The performance objectives of the Mamma Mia intervention are presented in the left column in Table 1. These are specifications of the overall goals and defines more clearly what it entails to prevent PPD in behavioral terms at the personal and interpersonal levels. It is, for instance, important that the mother regularly screens herself for depressive symptoms and is encouraged to seek help and support, and provided with immediate and additional on-screen support (ie, "just-in-time" therapy; see PO4 in Table 1). In addition, due to the already comprehensive approach to PPD in Mamma Mia, and the increasing complexity and additional costs associated with differentiation of subgroups, it was not feasible to differentiate the population at this stage in the development.

Overall, the needs assessment identified that for women to be able to prevent and alleviate perinatal depressive symptoms, they have to successfully manage the transition to parenthood. This also entails engaging in relationship- and health-promoting social and mental activities, both at a personal level and in relation to the woman's baby and her partner. The woman regularly needs to assess how she is doing and, if necessary, request help and support. For women who develop depressive symptoms, it is important that she is provided with immediate help and support to take the edge off the symptoms, as soon as possible, to prevent any adverse consequences as well as the onset of a more serious clinical depressive disorder or recurrent depressive episodes.

Personal and Interpersonal Determinants

Once the performance objectives were specified, we returned to the needs assessment to identify modifiable factors that in some way cause or can prevent perinatal depressive symptoms (see top row in Table 1). At the personal level, knowledge about or awareness of perinatal depressive symptoms is probably the first prerequisite for preventing depressive symptoms (see eg, PO4 in Table 1). Becoming aware of perinatal depression, the woman may need to adjust or relax certain expectations and attitudes that otherwise may nourish symptoms of depression (see eg, PO1 in Table 1), and become more tolerant and self-accepting of her pregnancy and becoming a parent. At the interpersonal level, it is important that the woman gets to know her baby and forms an emotional bond during pregnancy, as this predicts secure attachment 1 year postpartum [49] (see eg, PO2 in Table 1). This may help her establish an early relationship with her baby characterized by amazement and enjoyment rather than disruptive behaviors or a lack of contact between the mother and her child. Furthermore, by taking care of her partner relationship, this may act as a buffer against distress during pregnancy and the postpartum period, and be the first step in reaching out for help in cases where a woman may feel burdened or saddened (see eg, PO3 and PO5 in Table 1).

The Change Objectives

The next step was to develop and specify the change or learning objectives. The change objectives constitute the actions the mother has to do to carry out the performance objectives, and are a response to the question "What do intervention users need to change or learn to accomplish the performance objectives?" The performance objectives, determinants, and change objectives for the Mamma Mia intervention are summarized in Table 1. The cells in Table 1 thus constitute the building blocks or change processes in the intervention. This can be seen as an overview of the active ingredients of the intervention and as a blueprint of the theoretical treatment rationale.

In reviewing the literature and analyzing the problem of PPD, it became apparent that there is a great need for psychoeducational information (ie, knowledge) about postpartum depressive symptoms and the importance of the couple's relationship and emotional bonding between a mother and her child during pregnancy and the postpartum period. Many women report being unfamiliar with symptoms of depression during pregnancy and after childbirth, and express or hold certain expectations or beliefs that can be counterproductive with regard to one's mental health. For example, learning that a depressed mood usually makes people more socially withdrawn and quickly lose interest in social activities can result in a lack of interactivity and emotional bonding with the baby. This may cause some women to attribute failures to connect with the baby to personal and stable characteristics by themselves rather than realizing that most babies may need up to 10 seconds to respond to parent-initiated interaction. Thus, by learning simple attachment strategies such as "being with baby" and the principle of "wait, watch, and wonder," the woman may prevent the spiraling of such vicious transactional cycles.

In the wider context, this matrix of change objectives functioned as a backbone for the selection of theories and methods in the translation of these into the actual intervention.

Table 1. Performance and change objectives for the Mamma Mia intervention.

	Determinants			
Performance objectives	Knowledge	Expectancies and attitudes	Attachment, emotion regulation, and help seeking	Relationship satisfaction and communication skills
PO1: Cope adaptively with becoming a parent	K1.1: Understand that mixed feelings are normal postpartum	EA1.1: Accept that experiencing the maternity blues is normal	AEH1.1: Prepare friends and family for the expecting baby and upcoming life changes	RC1.1: Demonstrate the skill to effectively communicate and share needs and expectations toward partner
	K1.2: Acknowledge that detailed planning can be counterproductive	EA1.2: Let go of the need for rigorous and detailed planning and control		
	K1.3: Recognize that the postpartum period is hectic, and that it is important to be realistic about what one can achieve	EA1.3: Believe that breast-feeding is a skill that needs to be learned, and that there are alternative options		
	K1.4: Learn about alternatives to breast-feeding			
PO2: Engage in positive parent-infant interactions	K2.1: Understand how PND can interfere with bonding between a mother and her infant	EA2.1: Reflect confidence in parenting ability	AEH2.1: Experience “being with baby”	
	K2.2: Learn about infant development	EA2.2: Attribute failures to connect with infant to situational factors	AEH2.2: Identify and recognize the sleep-wake cycles of infants	
	K2.3: Become aware of the infant’s attention and communication skills	EA2.3: State that infants need time to react and respond	AEH.2.3: Demonstrate parent-child interaction and engage in appropriate attachment behaviors	
		EA2.4: Set realistic personal standards and expectations for the prepartum and postpartum period	AEH2.4: Utilize the principle of “wait, watch, and wonder” in interactions with her baby	
		EA2.5: Accept “good enough” parenting		
PO3: Engage in proactive and positive physical and mental activities	K3.1: Know the rationale for the positive psychological approach and learn the benefits of engaging in positive activities	EA3.1: Feel positive about involving the partner in preparations and taking charge	AEH3.1: Use techniques to enhance subjective well-being	RC3.1: Correctly perform exercises that can increase relationship satisfaction
	K3.2: Understand the pros of enhancing the partner relationship during pregnancy		AEH3.2: Practice relaxation and being present minded	RC3.2: Demonstrate more positive emotions toward partner while decreasing the expression of negative emotions
	K3.3: Understand that certain beliefs or assumptions about partner relationship are false or myths		AEH3.3: Make a list, plan, and engage in pleasant activities	RC3.3: Demonstrate a set of principles and use techniques for improving partner communication
			AEH3.4. Engage in physical activity	

	Determinants			
Performance objectives	Knowledge	Expectancies and attitudes	Attachment, emotion regulation, and help seeking	Relationship satisfaction and communication skills
PO4: Get help and support if depression is indicated	K4.1: Know that there are effective methods for managing depressive symptoms	EA4.1: Feel positive about and see the need to screen for depressive symptoms	AEH4.1: Ask for partner support	
	K4.2: Describe potential symptoms of postnatal depression		AEH4.2: Call mental health hotline	
	K4.3: Realize that social withdrawal from partner and others is a part of the problem		AEH4.3: Contact general physician	
			AEH4.4: Active and continued participation in Mamma Mia	
PO5: Cope adaptively with symptoms of depression	K5.1: Learn to identify certain maladaptive ways of thinking and behaving	EA5.1: Expect that using the techniques learned in Mamma Mia can be beneficial	AEH5.1: Learn a set of techniques to improve mood	See RC1.1 and RC3.1-3.3
		EA5.1: Feel positive about asking for help and support, and expect that it can be beneficial	AEH5.2: Change or replace ineffective mood strategies	
			See also change objectives for PO2 and change objectives AEH4.1-4.4	

Step 3: Theory-Based Methods and Practical Strategies

In the third step, the IM protocol addresses the selection of theoretical methods based on the change objectives identified in Step 2. Selected methods are then translated into practical strategies giving consideration into “what should be done” and “how it should be done” (ie, strategies for use). As seen in Table 2, both self-regulation and information processing theories served as the foundation for the development of practical strategies to influence knowledge, expectancies, and attitudes. These represent a conscious effort to become aware of and reflect on the physiological and psychological processes during pregnancy and after childbirth, and encourage a more flexible, open-minded attitude toward a range of beliefs and ways of thinking. Although knowledge or expectancies may not translate directly into behavior change, they provide a framework for understanding the importance of engaging in (self-)caring behaviors during the perinatal period, which may translate into behavior change (eg, finding alternatives to breast-feeding or selecting positive persons for support). In the remaining part, methods and strategies for use are presented and reorganized in accordance with the overall goals and determinants in the intervention from the user’s perspective.

Table 2. Mamma Mia determinants, methods, and strategies for use.

Determinants	Methods	Strategies for use
Knowledge	Consciousness raising (TTM[a])	Psychoeducation, guidelines, and recommendations are often followed by, for example, reflective questions intended to raise awareness about certain counterproductive expectancies or attitudes (eg, "I can't ask anybody for help, I should be able to take care of my own baby")
	Active learning (ELM[b] and SCT[c])	Psychoeducational information, cognitive and behavioral assignments, brief and many learning moments with repeated content over time to provide opportunities for rehearsal.
	Elaboration (ELM[b])	Information should be relevant, easily understandable, and rewarding to follow. Aligned with the chronology of the physiological and psychological processes during pregnancy and postpartum.
Expectancies and attitudes	Goals/personal standards (SRT[d])	Promote acceptance of a lesser need for detailed planning and rigorous control (eg, unexpected events may occur during birth).
	Normalization (NSI[e])	Normalize mixed feelings, potential failure to breast-feed, and feeling low on energy, which can assist with relaxation.
	Self- and environmental re-evaluation (TTM[a])	Stimulate appraisal to self-assess depressive symptoms, reinforce partner involvement, reinforce early parent-child bonding (eg, fantasy baby).
	Verbal persuasion (SCT[c])	Communicate optimism about users' parenting abilities, benefit of participating in Mamma Mia, and support-seeking behavior.
Attachment, emotion regulation, and help-seeking	Newborn Behavioral Observation	Video demonstrations of infant sleep-wake cycles and social interactive skills, homework assignments.
	Circle of security	Illustrated graphics to help parents understand their baby's needs and activate appropriate attachment behaviors.
	Positive psychotherapy (PPT[f])	Provide concrete tasks that focus users' attention on all the good things in life to enhance positive emotions, engagement, and a sense of meaning (eg, gratitude exercises); homework assignments.
	Mindfulness (PPT[f])	Audio-taped instructions that foster being in the moment, which are provided as downloadable audio files for personal use; homework assignments.
	Behavioral activation (PPT[f])	Recommendations for physical activity during pregnancy and after childbirth. Compile a list of pleasant activities and schedule pleasant activities over the course of the intervention.
	Stress and coping social support theory/relational regulation	Edinburgh Postnatal Depression Scale to assess depressive symptoms, encourage asking for partner support and/or general physician, provide a phone number to a mental health hotline.
	Metacognitive therapy	Audio-guided instructions and exercises (eg, attention training technique) to induce a state of awareness of internal events (eg, excessive worry) without responding cognitively, emotionally, or behaviorally; homework assignments.
Relationship satisfaction and communication skills	Gottman's method (couples therapy)	Couple exercises and homework to build closeness with partner, create a supportive relationship, and learn to manage conflicts (eg, softening technique); homework assignments.
	Prevention and relationship enhancement program	Video demonstrations of communication and problem-solving skills (eg, speaker-listener technique); homework assignments.
	Nonviolent communication	Practice distinguishing observations from interpretation of actions, identifying and expressing one's feelings and needs in a nondemanding way, and be given performance feedback.

[a]TTM = transtheoretical model

[b]ELM = elaboration likelihood model

[c]SCT = social cognitive theory

[d]SRT = self-regulation theory

[e]NSI = normative social influence

[f]PPT = positive psychotherapy

Depressive Symptoms

Methods and strategies addressing depressive symptoms mainly pertain to help seeking in Table 2 where the Edinburgh Postnatal Depression Scale (EPDS) was used, so that mothers can self-assess their depressive symptoms in the present intervention. The EPDS has been validated for Internet administration [50,51] and a systematic review shows that it reduces the probability

for postpartum depressive symptoms [52], especially in combination with supportive counseling [53]. Therefore, women who screen positive (ie, EPDS score ≥ 10) are provided with immediate help and support based on metacognitive therapy (MCT) [54]. In MCT, a client deals with the cognitive mechanisms (eg, worry or rumination) that lead to emotional problems, rather than the content of specific negative thoughts, feelings, or beliefs, as in cognitive therapy. MCT can be effective for PPD [55], and appears promising for delivery in a digital format considering that some techniques and exercises are administered as audio-taped files, even in face-to-face sessions. For women who screen negative, evidence suggests that *universal* interventions may have a preventive effect in postpartum women [56,57]. Thus, it is equally important to be concerned with well-being and the maintenance of healthy mental practices or habits, even among nondepressed women.

Maternal Subjective Well-Being

Methods for emotion regulation were approached from a positive psychological perspective (see Table 2). In general, a positive psychological approach is concerned with doing more of what is right or healthy (ie, prescriptions about the good life), rather than correcting what is wrong (ie, symptom substitution). These methods have been previously documented to have a positive effect on well-being among healthy, normal adults, as delivered by Internet [58]. It has been suggested that disseminating a broad range of positive psychology interventions (PPIs) online, tends to increase their use and effectiveness [59,60]. Multiple PPIs were thus drip fed throughout the current intervention, although the intervention conveys that users should not use all the PPIs constantly or simultaneously, but rather try each PPI and use those that they find personally most relevant and useful. Consequently, a diverse range of PPIs were administered in the present intervention such as mindfulness (eg, mindful breast-feeding) [61], gratitude (eg, 3 good things) [62], acts of kindness [63], and other exercises (for examples, see [64]). It should be noted that some of the included PPIs were designed for mastery of adverse situations (ie, expressive writing and cognitive restructuring; see eg, [65]) and that several of these exercises were also adapted to support the processes involved in strengthening the couple relationship (eg, giving your partner a compliment or savoring past, positive relationship experiences).

Partner Relationship

As presented in Table 2, the intervention draws upon principles from Gottman's method for healthy relationships [66,67] and the Prevention and Relationship Enhancement program [68] to strengthen the couple relationship. Both methods are concerned with the basic ways in which couples communicate and manage conflicts or problems (eg, avoiding criticism and listening actively). The present intervention thus includes methods for effective communication such as sharing expectations with one another, showing an interest in one's partner (eg, building love maps), expressing positive emotions (eg, "I know you love me when..."), reflecting about how one argues and how these may be perceived by the partner, softening start-ups in conflicts, and discussing problems (eg, speaker-listener technique). Finally, to support conflict management and help couples resolve problems, the 4 principles of nonviolent communication were taught to help users (1) distinguish the assessment of or feelings evoked by the action of others, (2) identify and express their feelings and (3) needs, and (4) convey requests in a noncoercive or demanding way [69].

Parental Sensitivities and Mother-Infant Relationship

To sensitize mothers to their infants' competencies and to promote the development of a healthy mother-infant relationship, the intervention includes several items and concepts from the Newborn Behavioral Observation (NBO) system [70] and circle of security (CoS) [71]. The NBO system is an infant-focused, relationship-based method that includes, in this intervention, demonstrating infants' sleep-wake cycles, and their abilities to respond to stress, self-regulate, and socialize. Concepts from the CoS intervention were used in a complementary manner to NBO, with a focus on parents and their ability to reflect on their own and their infant's behaviors, thoughts, and feelings, and respond appropriately to the infant's signals. In the current intervention, it includes demonstrating the concepts of a secure base and safe haven, which represent the baby's need for exploration and learning and its need for protection and comfort, respectively. The principle of "watch, wait, and wonder" is used to support parents in their work with the concepts in CoS [72], and it should be noted that the current intervention also focuses on attachment processes during pregnancy. Taken together, the current intervention focuses mainly on the social and interactive components of NBO and CoS where the goal is to support mothers learn to understand their infant and respond in a positive and developmentally supportive manner (Table 2).

Step 4: Program Components and Materials

Information and Communication Technology

Today, information and communication technology represents a promising channel for dissemination of Internet interventions in Norway. Most Norwegians have access to, and use, the Internet. On an average day, 83% of women in the population report having used the Internet; whereas 96% and 100% of women in childbearing age (ie, 16-44 years) reported having used email and the Internet in the last 3 months in 2014, respectively [13]. Given that many women in childbearing age use the Internet and are so-called eHealth information seekers as described earlier, the decision to develop an Internet intervention was taken at the very beginning of the process. One important aspect to this decision, however, is that 73% reported in 2013 to have access to mobile phones, and 61% of the population had access to tablets [73]. Thus, it was considered important to make the intervention platform independent (ie, independent of any specific hardware or software) to ease the accessibility and use of the program (ie, available on smartphones and tablets). The Internet was considered an appropriate and cost-effective medium for preventing PPD, especially given that the long-term goal is to offer the intervention to all pregnant women in Norway (ie, about 60,000 women/year) [74]. Furthermore, it was to be a fully automated self-help intervention, which is consistent with the aim of developing a primary preventive intervention for perinatal depressive symptoms.

Tunneled Design and Program Structure

The 2 core organizing principles for the delivery of intervention content are the tunneled information architecture and noise reduction (see Multimedia Appendix 1) [75]. The first principle, the tunneled design, entails that information is presented in a predetermined sequence page-by-page and session-by-session. It can have a negative effect on users' perceptions of efficiency, but seems to have a positive effect on intervention adherence and acquisition of knowledge [76]. The second principle, noise reduction, means that information only becomes available at the right time during pregnancy and postpartum rather than being available all the time. For example, in its simplest form, information about fetal development is delivered according to gestational week. This is done to avoid distractions and to minimize cognitive load on users [77]. Consequently, user navigation is, most often, limited to only "back" and "next" options. Several Internet interventions have used a tunneled design and reduction, demonstrating the feasibility and promise of such an information architecture for mental health and behavior change [78-81].

The intervention is used individually by the mother with health professionals, partners, or other actors only involved indirectly through the mother, but with no direct access to the program. For access to each session, users receive an email with a unique link. By clicking on the link, users are directed to a sequence of websites that are unique for that particular session. In the prenatal phase, emails with sessions are scheduled on a weekly basis, while sessions are scheduled 3 times a week in the active postpartum phase (ie, weeks 3-9). This is followed by a low-intensity phase with weekly and eventually monthly sessions. The intervention starts in gestational weeks 18-24 and lasts until the baby is about 6 months. In total, the intervention consists of 44 sessions over a period of 11.5 months. Each session is designed to take about 10 minutes and must be completed before users can access the next session. This is done to ensure that relevant information has been reviewed and to create continuity and a narrative in the program. If a mother discontinues a session, she is required to complete the previous session before she can move on to the current session. If the mother is on schedule, the session will end by directing her to the intervention home page. On average, a typical website during a session consists of 80-100 words and a typical user may have 10-15 pages per session (ie, the exact number of pages per session may vary due to tailoring). For a demonstration, see [82].

A session may utilize various functionality and interactivity to engage users, but typically consists of psychoeducational information, interactive tasks, and cognitive or behavioral homework assignments (for examples, see Multimedia Appendix 2). Psychoeducation is most often text-based information supplemented with printable documents, graphical illustrations, and video demonstrations. As an example, mothers are provided with descriptions of the infant's behavioral states followed by video demonstrations (see "Newborn Behavioral Observation" under "Methods" in Table 2). Interactive tasks typically include on-screen activities such as audio-guided instructions, quizzes, questionnaires, and written and reflective assignments. Examples of interactive tasks are filling in the EPDS questions and performing audio-guided mindfulness exercises (see "Self-and Environmental Re-evaluation" and "Mindfulness" in Table 2). Mindfulness involves experiential on-screen and homework exercises presented repeatedly. Mindfulness and re-evaluation represent a good example of how users are actively engaged in their change process where information is often elaborated and rehearsed over time (see "Active Learning" and "Elaboration" in Table 2). Homework assignments often accompany interactive tasks and are always related to the topic of a particular session. They may consist of reflecting upon why the woman reported to be feeling down lately and whether she needs someone to talk to, and practicing mindfulness in everyday life. A final example demonstrates how methods for the partner relationship are translated into the actual intervention; a mother is provided with psychoeducational text-based information about how conflicts may escalate and is allowed to watch a video demonstration of a couple going into an argument about trivial issues (eg, leaving dirty laundry on the bathroom floor). For homework, she is given a downloadable and printable document where she and her partner are supposed to discuss how they usually argue and how these conflicts are perceived. The lesson learned is that all couples argue from time to time but it is not the number of conflicts that determines whether the couple's relationship will be in a satisfying relationship or not, but rather how these conflicts are handled and perceived. Two sessions later, she is provided with a blueprint for the softening technique to learn to handle conflicts in a gentler and more appropriate manner—see "Gottman's Method (Couples Therapy)" and "Prevention and Relationship Enhancement Program" in Table 2.

At the end of each session, the user is directed to the intervention home page (see Multimedia Appendix 1). This home page contains an overview of the entire intervention where users may review completed sessions and retrieve all learning materials. The home page employs a top-down hierarchical information architecture and helps users find desired content by identifying the broad topics for each sessions. It is interesting that the tunneled design, on the one hand, restricts a user's navigational freedom, but may foster an interactive dialog that would otherwise be difficult to mimic or require more costly and early stage technology (eg, relational agents, semantic Web or natural language processing). On the other hand, the hierarchical design relaxes the restraints imposed by the tunneled design and allows users to freely explore and rehearse completed contents.

Personification and Tone-of-Voice

In its most rudimentary form, the Internet is a text-based medium. However, even static text material can mimic features of human-to-human dialog that can foster a sense of relationship or alliance between an intervention and the user [83-85]. This may, in turn, be positively related to the use and impact of the intervention [86,87]. A review by Kennedy and colleagues [88] found that users do have a preference for empathy and other relational behaviors in dialog systems for various health topics. Therefore, the intervention is embodied or personified by depicting 3 different persons alongside text and other interactive components, with each representing their special topic. The 3 persons are presented in 3 separate "rooms," which organize and structure the content and intervention materials: (1) the

mother's room, (2) the couple's room, and the (3) baby's room. The agent in the mother's room conveys information about the mother by screening for depressive symptoms and administering the well-being component, as described earlier. The 2 remaining agents provide information and exercises related to the couple's relationship and parental sensitivity, respectively.

The information is conveyed in a pleasant, but informal tone-of-voice without using any slang and becoming too friendly and overly humorous. Rather, the intervention provides users with social cues that, in turn, may elicit corresponding social responses. For example, in each session, one of the agents greets the user at the beginning of the session and says goodbye at the end, as if the intervention was a person. Several sessions also start by reviewing homework from the previous session and end by reminding the user about her homework for the next session. This may develop and build a user narrative with a past, present, and future that may help establish a form of continuity in the relationship. Oftentimes, personal pronouns were used and an active voice was preferred over a passive voice.

Step 5: Adoption and Implementation

The Norwegian Public Association for Women's Health [89] is a nonprofit organization that works toward improving women's living conditions. They have sponsored the development of the intervention, cover its operating costs, and have the ownership and distribution rights to Mamma Mia in Norway. The plan is to implement the intervention in 2 steps: First, in the current version of Mamma Mia, the intervention is marketed directly to pregnant women by advertisements and banners on the Internet, social media, newspapers and magazines, and leaflets in GP's offices and midwife services at hospitals and well-baby clinics. Expectant mothers can enroll for the intervention from a dedicated website. At enrolment, mothers register their email address and create a password, and start the intervention the following Monday. The intervention is meant to be used individually and is free of charge. An important aspect, in this first step, is also media coverage on television, radio, and the Internet, as well as publication of the research on Mamma Mia in scientific journals and conferences. The latter is particularly important in terms of engaging health personnel and building confidence in the intervention to establish Mamma Mia as part of the basic and supplementary education for health personnel.

The second, long-term step in the implementation of Mamma Mia is to develop guidelines for the implementation of the intervention by GPs, midwives, and PHNs working in well-baby clinics. This entails designing supplementary education for midwives and PHNs who are provided with in-depth knowledge about Mamma Mia and trained in the delivery of the intervention in practice (eg, skills training), which, importantly, will de facto turn Mamma Mia into a guided Internet intervention. This currently ongoing work will be offered as supplementary training at the Centre for Child and Adolescent Mental Health [90] in 2016. It is worthwhile, however, to note that the implementation of any intervention requires more than just training and supervision. The implementation guidelines, which are currently work in progress, will be based on Fixsen and colleagues' implementation components framework [91]. This means that the implementation guidelines will also specify the formal and informal qualifications required for midwives and PHNs for the supplementary training and supervision. This is referred to as the competency drivers in Fixsen and colleagues' implementation framework [91]. However, according to their framework, it is equally important to focus on organizational drivers such as evaluation of midwives' and PHNs' performance, decision-support systems for quality assurance and improvement of Mamma Mia, administrative support to ensure that leaders, policies, procedures, climate, and other structures are aligned with the needs of midwives and PHNs using Mamma Mia in their practice, and strategies to work with external systems to ensure that necessary resources required to support the sustainability of Mamma Mia over time in well-baby clinics are available (ie, system intervention).

Step 6: Evaluation

The feasibility of the intervention was demonstrated during piloting [20]. Most participants found Mamma Mia to be acceptable and user friendly, and instilling confidence in the intervention, especially for participants recruited through hospitals and well-baby clinics. In the feasibility study, we also identified several issues that needed improvements, some of which are included in the latest version of Mamma Mia (eg, information about expectations for the postpartum period and the baby's sleeping patterns). Not all improvements were implemented as they were not feasible at the time (eg, requiring a high level of tailoring that would require extensive re-designing of the program), but remain a part of the long-term plans for the future quality improvement of the intervention.

At the time of this writing, there is an ongoing randomized controlled trial (RCT), which aims to test the effectiveness of Mamma Mia delivered in addition to usual care (for protocol, see [92]). The control group will receive only treatment as usual during pregnancy and maternity care. In Norway, ordinary prenatal and postnatal care includes visits to GP, midwifery services at well-baby clinics and hospitals, and PHNs at well-baby clinics. According to the Norwegian guidelines for prenatal and postnatal care [93,94], this will typically include about 9 consultations during pregnancy and about 10 consultations during the 1st year of postnatal care. We hypothesize that women who receive Mamma Mia will have lower depressive symptoms postpartum than women in the control group, and higher levels of subjective well-being.

In parallel to the RCT, we are interviewing participants who have received the Mamma Mia intervention. The purpose is to assure the quality of the program and contribute strategically to its quality improvement to avoid that the intervention becomes obsolete given today's rapid technological advances. To this end, we currently use the modified strengths, weaknesses, opportunities, and threats (SWOT) format. The SWOT format does not impose any specific assumptions or themes on the interview nor specify any particular types of answers. Yet, the SWOT format provides a certain structure to the interview and participants' reflections along 2 dimensions (ie, positive-negative and past-future).

Finally, it becomes important to assess the supplementary training for midwifes and PHNs and implementation of Mamma

Mia at well-baby clinics. The supplementary training is considered an important way of strengthening the implementation by making midwifes and PHNs familiar with Mamma Mia and associating it with knowledge or themes, which, according to the national guidelines, midwifes and PHNs should cover during consultations with pregnant and postpartum women and their partners. The supplementary training is also designed so that midwives and PHNs work actively with several of the organizational drivers to embed Mamma Mia into their local well-baby clinic and secure managerial and organizational commitment to the program, as part of the course requirements. This will hopefully help Mamma Mia to become an integrated part of the ordinary health services, and to ensure the dissemination and sustainability of the program.

Discussion

Preliminary Findings

The aim of this paper was to provide a comprehensive overview of Mamma Mia and its development using the IM protocol. There are 2 overarching aims in IM: first, it serves as a tool in developing an intervention in a systematic manner. It links the phases of the intervention development to theory and empirical evidence, and thus ensures that decision making is based on sound logic. Second, the IM protocol makes the process of intervention development explicit and transparent. As such, the paper describes a fully automated Internet intervention (Mamma Mia) that is available on personal computers, tablets, and smartphones, and is also platform independent (ie, available on devices from Apple, Google, Windows, etc). The program consists of 3 phases: the first phase, which starts in the 2nd trimester, consists of 11 sessions. It starts in the gestational week 18-24, and ends in the gestational week 40 (ie, estimated due date). The active phase starts when the infant is 2-3 weeks old, and lasts for 6 weeks, with 3 sessions/week. The final phase is the follow-up phase, which consists of 10 sessions spread over an 18-week period. In total, the program consists of 44 sessions disseminated over a period of 11.5 months.

Users of the program progress through the intervention in a tunneled sequence with content centered around topics concerning coping with the transition to parenthood and signs of sadness or depression, engaging with the baby and one's partner, and engaging in self-caring behaviors. The tunneled design is in accordance with the preparation and psychological reorganization of becoming a mother (and father, [95]), and the topics are based on our needs assessment, which, in turn, shaped the overall goals of the intervention. The overall goals were used to derive the performance objectives which together with its determinants were used to specify the change objectives that guided the development of the intervention program further, in terms of both the selection of theory-based methods and the principles underlying the design of intervention strategies and materials. It describes why and how change methods derived from, for example, couples' therapy (eg, [96]), positive psychology (eg, [64]), and attachment theory (eg, [71]), were included in the final intervention. As such, it unravels the logic of the development of the current intervention by linking objectives, theories, and actual program materials and activities, and provides a blueprint of the intervention.

The adoption and implementation planning was largely in line with what is prescribed by the IM approach. Potential adopters and implementers of the intervention were included in the development, adoption, and implementation processes from the outset [20], as recommended [24]. However, the actual course of action deviates from the IM approach in that the evaluation of the intervention has focused primarily on the efficacy of the intervention, in terms of behavior change, at the expense of process evaluation. Thus far, this study does not present any evaluation data. Hence, we cannot make any conclusions about its ability to prevent cases of depression, effectiveness in reducing depressive symptoms, or increasing subjective well-being. However, the transparency of the intervention content may assist with interpreting results from upcoming studies, anticipate any strengths and weaknesses with the intervention, and make potential changes or improvements that may enhance its effectiveness.

Generalizability

Mapping the development and contents of an intervention, as in this study, is useful and serves several purposes; first, it allows intervention designers to faithfully replicate effective interventions or attempt to design interventions that are even more effective. Second, it allows researchers to identify mechanisms or techniques contributing to intervention effectiveness (ie, *why* and *how* they work) and extend the evaluation of an intervention to, for example, other populations or settings (eg, for *whom* they work). Third, it may assist educational institutions and other agencies in developing training and implementation strategies for the intervention. Finally, health personnel may more easily determine whether interventions such as Mamma Mia may be an appropriate intervention for their practice and users. Therefore, we argue that this paper illustrates why such post hoc descriptions are useful and comply with the call for transparency and thorough descriptions of interventions that had been noted by numerous authors (see eg, [97-101]).

Systematic and comprehensive descriptions of interventions are necessary to ensure that we can describe aspects of interventions that are generalizable and identify determinants used to achieve change and how the use of theory can be translated into practical strategies. This is necessary to promote and deepen our understanding of the development of complex interventions and move on from general remarks and conclusions such as "...more extensive use of theory was associated with increases in effect size..." [102] to a higher level of specificity. More research is needed to identify exactly which theoretical frameworks and practical strategies work best. However, this is the stepping stone that can help advance the accumulation of scientific knowledge in eHealth and prevent "type III error," that is, rejection of an intervention's effectiveness because the intervention itself is poorly designed or implemented [103]. Furthermore, this may help advance the usefulness of systematic reviews, which identify, critically appraise, and synthesize relevant research, but this depends on the quality of the descriptions of the available interventions under investigation.

Therefore, we argue that researchers should routinely publish peer-reviewed intervention protocols separately prior to publishing results from evaluation studies (ie, similar to registration of trial protocols in registries such as ClinicalTrials.gov or International Standard Randomised Controlled Trial Number). This will help get a fuller understanding of what exactly is being evaluated, compare treatments against each other, and learn from previous development efforts.

Limitations

Based on the IM protocol, this paper has 1 key limitation: it was applied in a post hoc manner. Hence, the development process described in this paper deviates somewhat from the IM protocol. Nevertheless, it is important to note that the IM framework influenced the development of Mamma Mia, which makes a post hoc analysis both feasible and informative. For instance, a long-term perspective in this project made it possible for us to perform studies on the determinants of PPD in Norwegian mothers [33,34,104], which is a key step in intervention development according to IM. As previous studies illustrate, a retrospective IM-based analysis can be useful as it may point toward weaknesses in the intervention development process and the intervention itself [27,28], thereby providing information on any potential threats and issues that may arise mainly during the implementation and evaluation.

In a sense, this may also be one of the strengths of applying IM analysis post hoc and highlights why it is better to perform IM after the development of an intervention, rather than never. For example, dropout rates in Internet interventions tend to be high [105,106]. Thus, designing an intervention consisting of 44 sessions over a period of almost 1 year adds numerous opportunities for discontinuation. Although many pregnant and postpartum women may be "eHealth information seekers," it cannot be expected that this alone will sustain engagement with the intervention for extended periods, without any human involvement. By contrast, the lack of segmentation of the target population may prove to be more or less acceptable to certain subgroups. The intervention targets both young, first-time mothers who may feel more insecure and in need of support, and older, multiparous women who may have a greater sense of security and confidence in their parenting abilities. Mamma Mia may also be more appropriate for women living with a partner than single mothers and for women without complications. These factors may be used to guide future quality improvement of the intervention; however, currently, they also pose a threat to intervention effectiveness.

Conclusion

In the development of any type of intervention, intervention designers translate theory into practice. However, there are many steps during development where the theoretical methods believed to be effective can be "lost in translation." By applying the IM protocol to Mamma Mia, we have described the rationale and contents of the intervention, and opened up the black box for intervention designers, researchers, educational institutions, and health personnel working with perinatal mental health. The IM analysis of Mamma Mia helps to make clear decisions regarding intervention development from theory, empirical findings, and practical strategies that may contribute to its overall effectiveness. It also makes transparent some of the potential pitfalls such as its duration and lack of segmentation that may jeopardize intervention effectiveness. Overall, this paper adds to the plea for systematic reporting of intervention protocols that document the design and development of interventions, and accumulation of knowledge about interventions in the field of eHealth.

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Conflicts of Interest

FD was employed by Changetech AS at the time Mamma Mia was developed for The Norwegian Association for Women's Health (NKS).

Multimedia Appendix 1

Intervention screenshots from Mamma Mia.

[PDF File (Adobe PDF File), 2MB - resprot_v4i4e120_app1.pdf]

Multimedia Appendix 2

Transcripts from program materials.

[PDF File (Adobe PDF File), 413KB - resprot_v4i4e120_app2.pdf]

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Abbreviations

CoS: circle of security
EPDS: Edinburgh Postnatal Depression Scale
GP: general physician
IM: intervention mapping
MCT: metacognitive therapy
NBO: newborn behavioral observation
PHNs: public health nurses
PPD: postpartum depression
PPIs: positive psychology interventions
SWOT: strengths, weaknesses, opportunities, and threats

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Original Paper

TRAK App Suite: A Web-Based Intervention for Delivering Standard Care for the Rehabilitation of Knee Conditions

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Abstract

Background: Standard care for the rehabilitation of knee conditions involves exercise programs and information provision. Current methods of rehabilitation delivery struggle to keep up with large volumes of patients and the length of treatment required to maximize the recovery. Therefore, the development of novel interventions to support self-management is strongly recommended. Such interventions need to include information provision, goal setting, monitoring, feedback, and support groups, but the most effective methods of their delivery are poorly understood. The Internet provides a medium for intervention delivery with considerable potential for meeting these needs.

Objective: The objective of this study was to demonstrate the feasibility of a Web-based app and to conduct a preliminary review of its practicability as part of a complex medical intervention in the rehabilitation of knee disorders. This paper describes the development, implementation, and usability of such an app.

Methods: An interdisciplinary team of health care professionals and researchers, computer scientists, and app developers developed the TRAK app suite. The key functionality of the app includes information provision, a three-step exercise program based on a standard care for the rehabilitation of knee conditions, self-monitoring with visual feedback, and a virtual support group. There were two types of stakeholders (patients and physiotherapists) that were recruited for the usability study. The usability questionnaire was used to collect both qualitative and quantitative information on computer and Internet usage, task completion, and subjective user preferences.

Results: A total of 16 patients and 15 physiotherapists participated in the usability study. Based on the System Usability Scale, the TRAK app has higher perceived usability than 70% of systems. Both patients and physiotherapists agreed that the given Web-based approach would facilitate communication, provide information, help recall information, improve understanding, enable exercise progression, and support self-management in general. The Web app was found to be easy to use and user satisfaction was very high. The TRAK app suite can be accessed at http://apps.facebook.com/kneetrak/.

Conclusions: The usability study suggests that a Web-based intervention is feasible and acceptable in supporting self-management of knee conditions.

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KEYWORDS

internet; social media; web applications; mobile applications; usability testing; knee; rehabilitation; exercise; self-management

Introduction

Management of Knee Conditions

Musculoskeletal conditions are the second largest contributor to years lived with disability [1]. In the United Kingdom, a total of 33% of individuals aged 45 and over have sought treatment for osteoarthritis, with the knee being the most commonly affected joint [2]. The incidence of acute knee injuries is reported to be at a rate of 2.29 per 1000 in US population [3]. In the Netherlands, 45-55% of acute knee injuries develop into a long-term medical condition [4]. Patients may still experience movement deficiency 1 year following knee surgery [5]. In particular, participation restrictions may persist 2 years following total knee replacement [6].

To support the recovery or management of a long-term musculoskeletal condition, physiotherapy rehabilitation is typically recommended. Standard care for the rehabilitation of knee conditions involves exercise programs and information provision [7,8]. However, current methods of rehabilitation delivery struggle to keep up with large volumes of patients and the length of treatment required to maximize the recovery. Therefore, the development of novel interventions to support self-management is strongly recommended. Such interventions need to include information provision, goal setting, monitoring, feedback, and support groups, but the most effective methods of their delivery are poorly understood and require further research [9,10]. Finally, treatments need to be personalized, that is, targeted at individual needs, to improve prospects of rehabilitation [10].

Web-Based Intervention Approaches

Some argue that the Web-based approaches are more accessible, more convenient for patients, and can help counteract the current shortage of skilled therapists [11]. They can also be more effective in acquiring declarative knowledge [12]. Focusing on therapies that addressed deficiencies in patient knowledge and self-management skills, Web-based approaches were found to be more effective in increasing participation and exercise time [13]. In the context of physical activity interventions, it was found that over half of the controlled trials of Web-based interventions reported positive behavioral outcomes [14]. Similarly, findings of a systematic review demonstrated the effectiveness of Web-based patient education interventions, that is - interventions that are associated with producing changes in self-care behavior, on patient outcomes [15]. A meta-analysis of Web-based cognitive behavioral interventions demonstrated a small effect when using pain scale as the main outcome in comparison to waiting list control groups [16]. These results indicate the potential of Web-based interventions as a therapeutic tool for chronic pain associated with decreasing treatment costs and side effects. In general, Web-based interventions in patients with somatic diseases were found to be effective/cost effective, or at least promising in this regard [17]. In particular, Web-based physical activity interventions proved to be more effective than a waiting-list strategy [18].

Although Web-based behavioral interventions can be effective, poor adherence is commonly associated with such interventions [19]. The differences in technology and interaction, rather than the health care area itself, were found to be good predictors of adherence. The most effective Web-based interventions are interactive and flexible, thereby allowing patients to select information that is of relevance to them, and their learning at a particular point in time [15]. In addition, social media offer an opportunity to improve the efficiency and effectiveness of health care by providing an alternative mechanism to facilitate self-management of chronic diseases [20].

The Health Care Industry and the Web

"Social media" is a term used to refer to a group of Web-based apps that allow the creation and exchange of user-generated content [21,22], such as social networks, forums, blogs. To reach stakeholders, leverage collaboration, and personalize care, the potential of social media needs to be explored [23-25]. They have already demonstrated a rising influence on patients' decision-making process (eg, what services to seek out, which doctor to consult). Not surprisingly, it has been suggested that social media will inevitably be an integral part of the future landscape of health care [26]. However, the accent here is on the word "future," because the health care industry is currently lagging behind in its use of social media to communicate with patients. For example, a study of social media use among Fortune 100 companies revealed that health care was the least active industry in this area [27]. Similarly slow adoption rates have been reported in the United Kingdom [28]. This tendency may be explained by a combination of various factors.

Health Care Industry Factors for Web Resistance

Publicly financed health care organizations such as the National Health Service (NHS) have traditionally been risk averse. Subject to budgetary constraints, they strive to minimize the possibility of failure. Therefore, they tend to only adopt widely accepted, proven technologies [29].

There is a lack of best practices and robust metrics to measure success of investments in social media and produce reliable return-on-investment figures. Although there are several surveys, case studies, trials, and working examples, systematic evidence on the clinical outcomes of social-media-enabled health interventions is yet insufficient [23].

There is a general lack of trust. Social media are rife with misinformation. In heavily standardized and regulatory-driven industries such as health care, it is imperative to provide well-recognized and accredited sources of information [30].

Privacy, security, confidentiality, and liability questions present further concerns [25]. Communication between health care providers and patients must comply with current data security and privacy legislation [31,32]. Because of increased privacy laws and regulation, health care-centric discussions need greater moderation than discussions related to any other industry.

Health professionals tend to link the use of social media to deprofessionalization of their expertise, and they believe that it can undermine the traditional doctor-patient relationship [33].

Clearly, there are numerous hurdles to overcome, and engaging with social media can incur considerable risks. Despite their increasing popularity, their uncontrolled nature raises serious concerns about the quality, reliability, and accuracy of health information. The benefits health care may gain from investing into this relatively new technology are still fairly unclear [34]. Nonetheless, it provides a great opportunity to improve the quality of communication between patients and health care professionals, thus potentially improving health outcomes. In particular, the emergence of social networking sites (SNSs) created entirely new ways of social interactions. The traditional vertical dissemination has been replaced with a horizontal flow of information. Social networks dedicated to patients function as virtual support groups, which can offer a wide range of information and support needs that may not be met as part of conventional health care [35]. For example, the website "PatientsLikeMe" [36] was designed to facilitate conversation between patients suffering from the same medical condition. Another example is CureTogether [37], which helps its users to anonymously track the progress of conditions, collect data to aid research, and help patients better understand their bodies.

The majority of Web-based physical activity interventions focused on health promotion [38,39], often in a specific medical context such as obesity [40], diabetes, [41], or aging [42], but very few exist in areas such as osteoarthritis [43] or rheumatoid arthritis [44], where physical therapy is recommended. To our best knowledge, there is no evidence of a successful interactive app for patients with acute knee conditions.

Methods

Primary Aim of the Study

The primary aim of this study was to demonstrate the feasibility of a Web-based, interactive app, and to conduct a preliminary review of its practicability as part of a complex medical intervention in the rehabilitation of knee disorders. Based on the precognitions of stakeholder expectations, our objective was to examine possible ways of facilitating remote self-care. Ethical approval for this study has been granted from the National Research Ethics Service via the South East Wales Research Ethics Committee Panel (ref 10/MRE0928).

Choosing the Platform

Incorporating the social networking aspect into the proposed app is a critical success factor. There are 2 ways to accomplish this: (1) by organizing a new community from scratch or (2) carving out a chunk from the audience of the mainstream SNSs. Open source social networking platforms such as Elgg [45], Oxwall [46], or BuddyPress [47] provide an out-of-the-box solution for building new online societies. A disadvantage of this approach is that users need to register and remember their login credentials to yet another account and learn how to effectively use a new software environment. Alternatively, Facebook Login [48], Twitter OAuth [49], Yahoo OAuth [50], Google OAuth 2.0 [51], and Disqus [52] offer ways to integrate third-party websites with social media by allowing authentication and content authoring from external websites. Additionally, Facebook allows developers to host their apps on Facebook, which allows effortless promotion via "likes" and recommendations as well as contextual advertising. A wealth of user information (name, age, interests, groups, friends, etc) comes for free and users are faced with a familiar interface, which requires no special training, thus, not incurring a significant learning overhead. A Facebook app requires no installation, is platform neutral in terms of operating systems and Web browsers, and is, in principle, accessible from any Internet-enabled device. The interactive nature of Facebook and communication with other users makes it straightforward to integrate a virtual support group into the app. However, such communication together with the collection of user information may raise privacy concerns. This means that, in addition to Facebook's own privacy policy [53], it needs to be made clear to the users how their information will be used within the app. From developers' perspective, the negative consequences of choosing Facebook as a platform include loosing independence and developing to interfaces that may change, possibly breaking the app.

In the context of self-management interventions, Facebook seems like a suitable platform due to its widespread usage. In the United Kingdom, 61.1% of Internet users use Facebook at least monthly [54]. Similarly, in the United States, 74% of adult Internet users use SNSs, with Facebook being the preferred choice (71% of adult users) [55]. The percentage of Internet users within a specific age group who use Facebook is high in both young and middle-aged adults: 86% of users are in the age group of 18-29 years, 73% in the age group of 30-49 years, and 57% in the age group of 50-64 years [56]. Overall, Facebook users in the United States are currently distributed across different age groups as follows: 13-17 years (5.4%), 18-24 years (23.3%), 25-34 years (24.4%), 35-54 years (31.1%), and over 55 years (15.6%), with the 3 older groups recording a growth of 32.6%, 41.4%, and 80.4%, respectively, within the last 3 years [57]. This trend is contrary to popular belief that Facebook is mostly used by teenagers. When it comes to gender, the distribution of Facebook users is fairly balanced in comparison to other SNSs; there are slightly more female (53.3%) than male (45.6%) users. On the other side, acute knee injuries are most prevalent in young and middle-age adults (75.6% of knee injuries occur in men with a mean age of 32.9 years, compared with 24.4% in women with a mean age of 35.3 years) [58], whereas the percentage of individuals treated for long-term conditions such as osteoarthritis is 31% women and 23% men within the 45-64 age group and 44% women and 35% men within the 65-74 age group [2]. These findings indicate that a self-care app delivered via Facebook may have potential health benefits for a high percentage of adult users. For these reasons, we decided to use Facebook as the main platform to develop a Web-based app for self-management of knee conditions.

However, there is evidence that engagement with physical activity interventions delivered using websites tends to be low unless concerted effort is taken to address the issue [13]. Engagement can be increased by focusing on self-regulation through the use of modern devices such as mobile phones [39].

Mobile phones are globally more prevalent than computers for Internet access [59]. In addition, they are designed to be portable. For these reasons, mobile phones may provide a better platform than large screen devices (eg, desktop, laptop, or tablet) to increase engagement in the context of an exercise-based rehabilitation program. While the overall Web-based app may be too complex to be delivered efficiently on a relatively small mobile phone screen, the self-monitoring aspect of its exercise component is ideally suited for this medium. Therefore, multiple platforms are required to maximize both accessibility and engagement in a Web-based app for self-management of knee conditions. We chose to implement a Web-based intervention as a suite of apps in which a Facebook app is complemented by 2 native mobile apps.

Needs Assessment

To ensure that the final product is optimized for its intended audience in terms of user needs, we conducted a meta-analysis of publications concerning the rehabilitation of musculoskeletal conditions to shed light on patients' information needs and self-care support requirements. Approximately 100 titles were screened and 20 of them were considered for inclusion. We conducted content analysis to group information into themes. In terms of supporting self-care, we identified the following 5 key areas: (1) sufficient and comprehensible information provision about generic and condition-specific matters; (2) tailored exercise plans that take patients' individual circumstances into account; (3) recovery monitoring based on data supplied by patients (functional test and questionnaire surveys); (4) a virtual community of patients; and (5) support for patient-doctor interactions.

Goal Analysis

The goals defined in the needs assessment phase were broken down into component tasks by asking questions such as “In order to achieve this goal, what does the user need to know or be able to do?” Tasks were then structured around scenarios of interest and in the context of specific types of users [60]. User stories were used to document user requirements in a quick, informal way. A prioritized set of the most important user stories in our case are presented in Textbox 1.

Textbox 1. Prioritized set of the most important user stories.

- As a patient, I want access to both general and condition-specific information.
- As a patient, I want to fill out questionnaires and functional tests so that I can track my progress.
- As a patient, I want to receive notifications about upcoming tests.
- As a patient, I want to see my results and compare them to other patients' averages.
- As a patient, I want visual feedback on my progress.
- As a patient, I want to know what tests I will have to do and when.
- As a patient, I want an exercise plan with detailed exercise descriptions.
- As a patient, I want to be able to talk to clinicians.
- As a patient, I want to start a new topic, post to existing ones, and comment and like other posts.
- As a patient, I want to be able to register.
- As a patient, I want to specify my demographic and medical details so that I get personalized content.
- As a patient, I want to be able to delete my account.
- As a patient, I want help on how to use the system.
- As a patient, I want a frequently asked questions (FAQs) section.

Information Organization Scheme

An information organization scheme defines the shared characteristics of content items and influences the logical grouping of those items [61]. In the context of Web projects, it is concerned with conceptually organizing information into groups (windows, pages, tabs, and other elements of user interface) and assigning names to those elements. The organization of information is a critical success factor; if users do not understand the scheme, they will not be able to find what they are looking for regardless of how easy it is to navigate the website. We chose to implement a task-based organization scheme based on an assumption that a user will typically use the system to perform certain tasks. By extracting these tasks from user stories and grouping them together into components, we obtained a coarse-grained structural model of the required system, through which the following 5 main modules were identified: Home page, Knowledge Base, Recovery Tracker, My Self-Care Plan, and Support Group.

The Home page was envisioned to display a welcome message with some basic information about the app. The Knowledge Base was intended to serve information provision purposes. The Recovery Tracker component is meant to be an interactive tool that helps registered patients assess their recovery progress. My Self-Care Plan contains information about the self-care plan throughout typical stages of rehabilitation. In relation to My Self-Care Plan, we specifically wanted to improve accessibility to information about exercises. Therefore, we designed a mobile app to allow a user to easily access and record information about exercises when outdoors or in the gym. Finally, the Support Group should provide a venue for patients to share their

experiences. The following section describes these modules and their relations in more detail.

Implementation

We developed the Taxonomy for RehAbilitation of Knee conditions (TRAK) app using extreme prototyping, a relatively new, iterative architectural approach, specifically designed for developing hypermedia apps in terms of increasingly functional prototypes [62]. Evaluation takes place at the end of each phase and prototypes are reviewed until the minimum requirements of acceptance have been met. Figure 1 shows a sitemap of the final version of the TRAK app suite. Different colors are used to distinguish between the modules whose names are listed directly under the Home page.

The Knowledge Base was intended to serve information provision purposes. Its content is grouped into 3 categories based on the results of a UK-wide survey of physiotherapists, with a specialist interest in knee conditions, regarding the types of information and advice they provide to patients [8]. The content of the 3 categories is displayed in the corresponding tabs. The "Physiotherapy" tab (Figure 2) contains articles whose purpose is to manage patients' expectations by providing information about the aims of physiotherapy, types of rehabilitation, rehabilitation goals, etc. The "Knee" tab provides information aimed at improving patients' understanding about the nature of the problem, for example, anatomy of the knee, different types of knee conditions, and the related symptoms. To get the necessary information across, our content needed to be easy to comprehend for patients with any level of health literacy and richly illustrated with graphics to reinforce understanding. Moreover, articles had to be compelling, original work, based on the best available evidence. Finally, the "External Links" tab was designed to supplement the original articles with a set of relevant links to credible external resources such as World Health Organization global recommendations on physical activity for health and Arthritis Research UK.

Figure 1. Sitemap of the Facebook app.

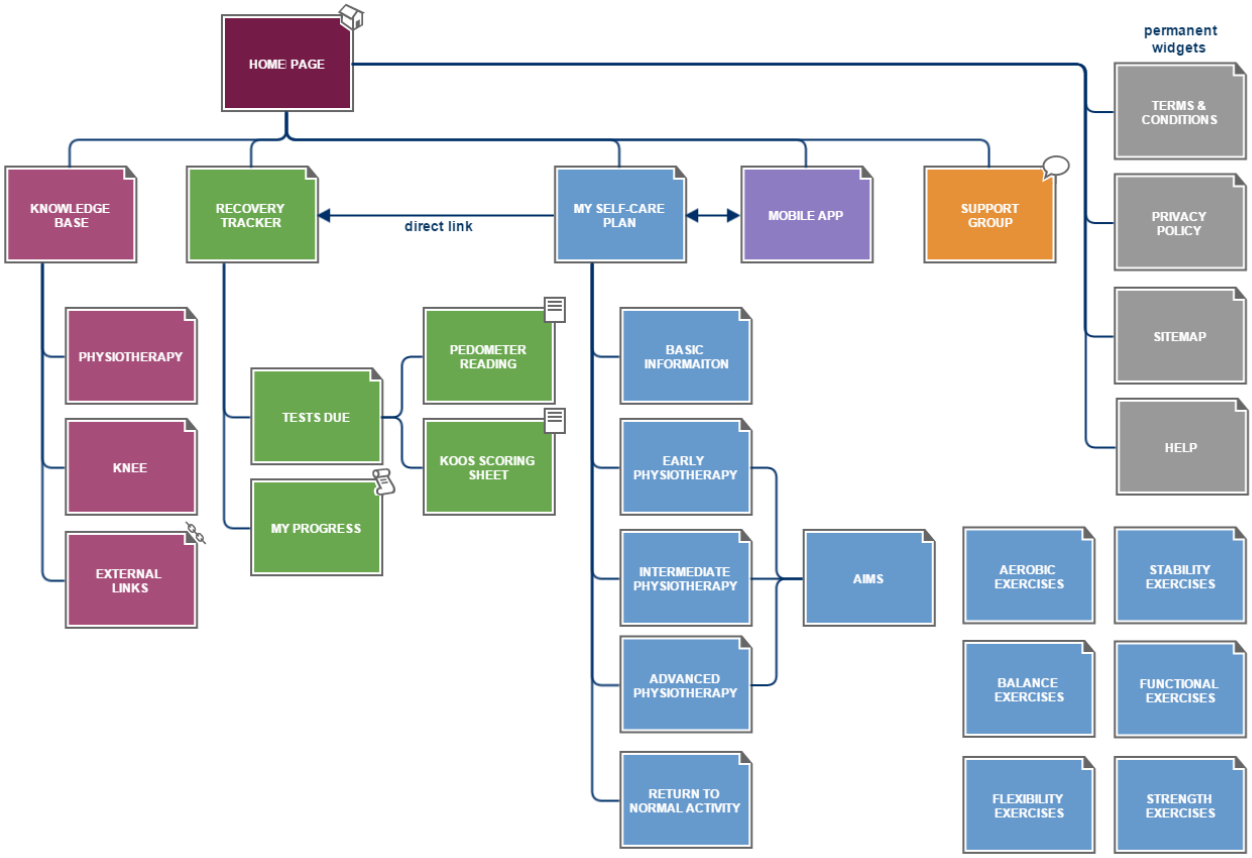
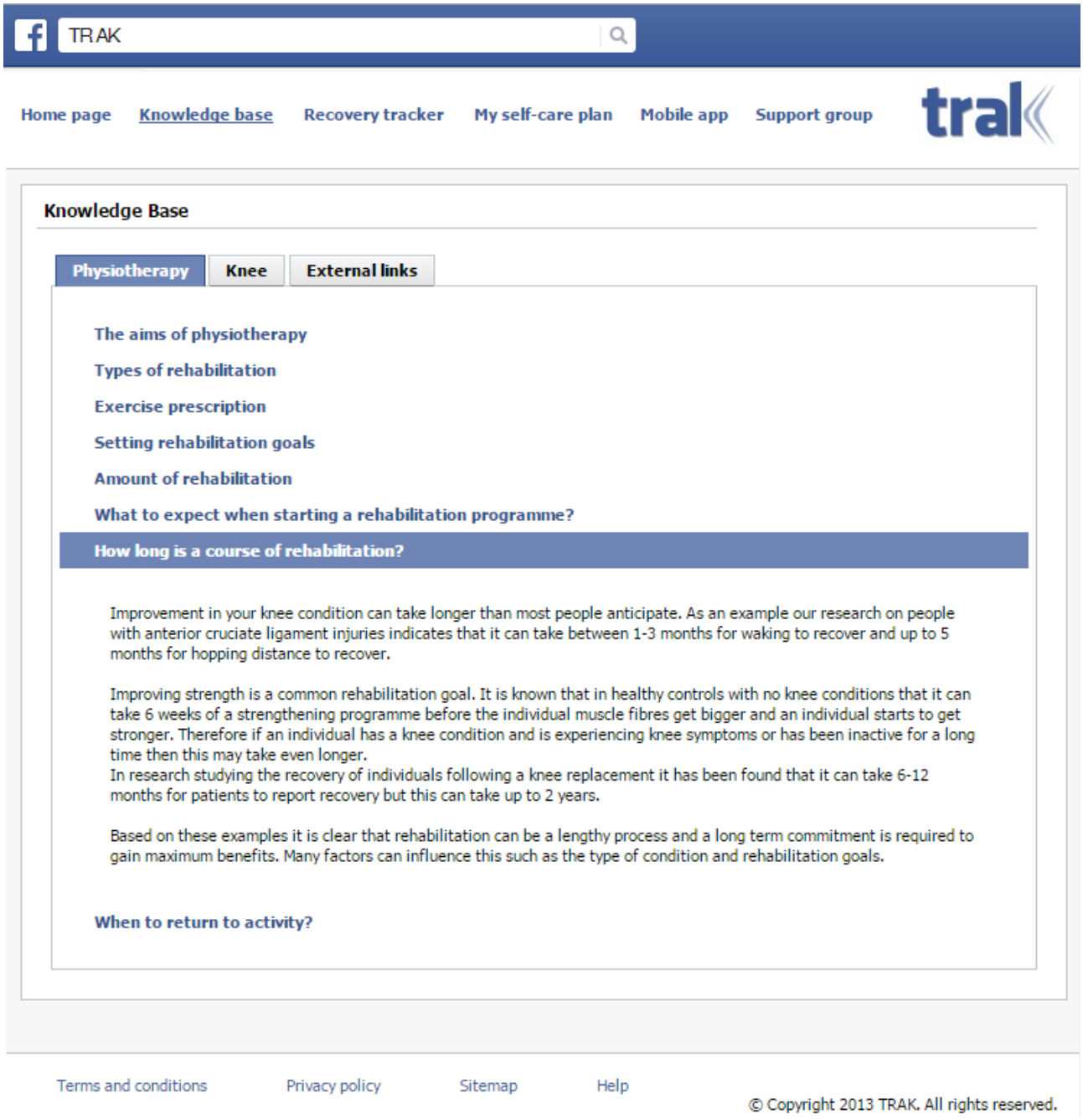


Figure 2. Knowledge base - Physiotherapy tab.



Recovery Tracker

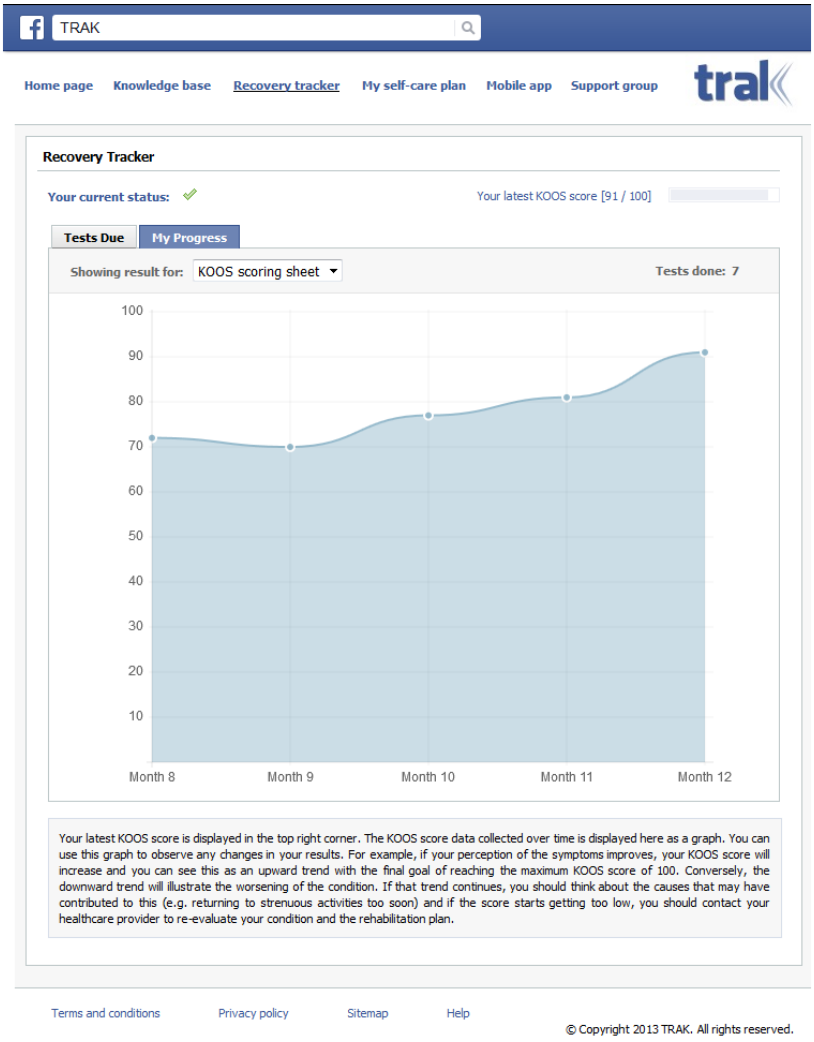
The Recovery Tracker component provides access to an interactive tool designed to help registered patients assess their recovery progress over the course of rehabilitation. It provides a schedule for collecting patient-reported outcome measures in the “Tests Due” tab, together with a checkbox to allow users to set email notification preferences. There are 2 types of patient-reported outcomes that are currently collected: subjective and objective. The subjective test is based on the Knee Injury and Osteoarthritis Outcome Score (KOOS) [63], a 42-item self-administered assessment of 5 outcomes (pain; other symptoms; activities of daily living; sport and recreation activities; and knee-related quality of life). This information is meant to help patients keep track of how they feel about their knee and how well they are able to perform their usual activities.

In addition to KOOS, pedometers are commonly utilized as part of knee rehabilitation as a way of increasing physical activity levels and allowing patients to objectively assess their performance [64]. Both tests (ie, KOOS and pedometer readings) are scheduled periodically. Taking into account the complexity of the KOOS survey, and in order to allow sufficient time for significant improvement to occur, the KOOS test is scheduled monthly. Pedometers record data on a daily basis, but most are nowadays equipped with a 7-day memory function. In order for the app to be less obtrusive, the pedometer readings are collected on a weekly rather than a daily basis.

Once a patient completes a test, the system evaluates answers and computes a test score, which is compared with previous scores and the average score of other patients in the same category and stage of rehabilitation to provide the patient with

feedback about their recovery. In addition to the latest test result, the patient can also track their progress over time. Namely, the "My Progress" tab provides patients with a summary of previous results (Figure 3). To simplify the interpretation of numerical information, a visual representation is provided in the form of a plot, with time expressed in weeks on the horizontal axis and test score on the vertical axis. Additional information is provided to aid interpretation of visual feedback, whose purpose is not only to inform a patient about their progress, but also to motivate them by relative improvement over time as well as to set realistic expectations by direct comparison to the average progress of other patients in similar circumstances.

Figure 3. Recovery tracker - My Progress tab.



My Self-Care Plan

The overall goals of My Self-Care Plan are to help patients to control joint pain and/or swelling, regain normal knee flexion and extension, regain a normal gait pattern and stability, regain normal muscle strength, regain normal proprioception, balance and coordination for desired activities, and achieve optimal functional outcome. To support these outcomes effectively, My Self-Care Plan contains an exercise-based rehabilitation program divided into 3 phases (early, intermediate, and advanced physiotherapy) followed by the final phase (return to normal activity) [65].

The length of the rehabilitation program will depend on patients' individual circumstances, and therefore, the proposed timelines are merely a guide. Exercise progression depends on completion of the current phase, before advancing to the next one. Each phase provides information about its own aims, exercise program, and progression criteria. For example, to proceed to Phase 3 (advanced physiotherapy), patients should meet the following criteria: (1) there should ideally be no swelling of the knee, but minimal swelling is acceptable; (2) comfortable walking; (3) full range of motion; and (4) symmetrical muscle strength. In addition to these criteria, Recovery Tracker can assist patients in deciding whether they are recovering sufficiently or not. When ready to progress, the next phase of physiotherapy can be selected from a drop-down menu (Figure 4). By default, the exercises shown when selecting My Self-Care Plan belong to the current phase.

Each rehabilitation phase is supplemented with information about its aims, advice, and a reference to Recovery Tracker. The exercises associated with each phase are divided into 6 groups, namely, aerobic, balance, stability, flexibility, functional, and strength exercises, which are accessible via the corresponding tabs. Each tab is populated with specific exercises appropriate for the given phase. Each exercise comes with a collapsible/expandable illustration section, which contains an

image and a short description (Figure 4). The exercise information originates from the TRAK ontology, which has been specifically designed to formally model standard care for the rehabilitation of knee conditions [8]. It incorporates over 100 exercises organized hierarchically (Figure 5). These exercises were selected through a systematic literature review [7] and a UK-wide survey of clinical practice. The TRAK ontology is encoded in the OBO flat file format, version 1 [66], which makes it machine readable and thus reusable in a variety of informatics apps. In other words, the content of the TRAK ontology can be automatically imported where needed. Textbox 2 provides an example of an exercise represented in the ontology. Such representation provides a unique identifier for each exercise, its preferred name, together with any other synonyms, definition, cross-references to external sources (eg, UMLS), classification using “is-a” relationship, and named relationships to other relevant concepts in the ontology.

Textbox 2. An example of exercise description in the TRAK ontology.

[Term]

id: TRAK:0000405

name: backward lunges

def: "Start position: Stand upright. Action: Step backwards with the affected leg and bend the affected knee until it is flexed 90 degrees, then slowly straighten up keeping the body weight on the affected leg and step back with the unaffected leg." []

synonym: "reverse lunges" EXACT []

xref: UMLS_CUI:C2019591

is_a: TRAK:0000135 ! lunges

relationship: performs TRAK:0000157 ! concentric contraction

relationship: performs TRAK:0000158 ! eccentric contraction

In relation to My Self-Care Plan, we specifically wanted to improve accessibility to information about exercises in order to better engage patients in the self-care program. Therefore, we designed a native mobile app to allow a user to easily access and record information about exercises without having to rely on an Internet connection, which may not be available in typical exercise environments, for example, outdoors or in the gym. Namely, My Self-Care Plan provides a selection of appropriate exercises as the patients progress through the 3 rehabilitation phases, while the mobile app allows them to keep a diary of exercise activities as they actually do them at home or somewhere else. The mobile app's functionality includes exercise selection, access to exercise instructions with an image, logging an exercise together with pain and effort required for its completion as well as any other comments, and tracking progress by monitoring pain and effort over time (Figure 6). As before, all exercise-related information was reused from the TRAK ontology. Any changes to the ontology stored on a cloud server are propagated automatically, thus allowing the mobile app to evolve seamlessly.

Figure 4. A description of a flexibility exercise recommended in Phase 2 (Intermediate physiotherapy).

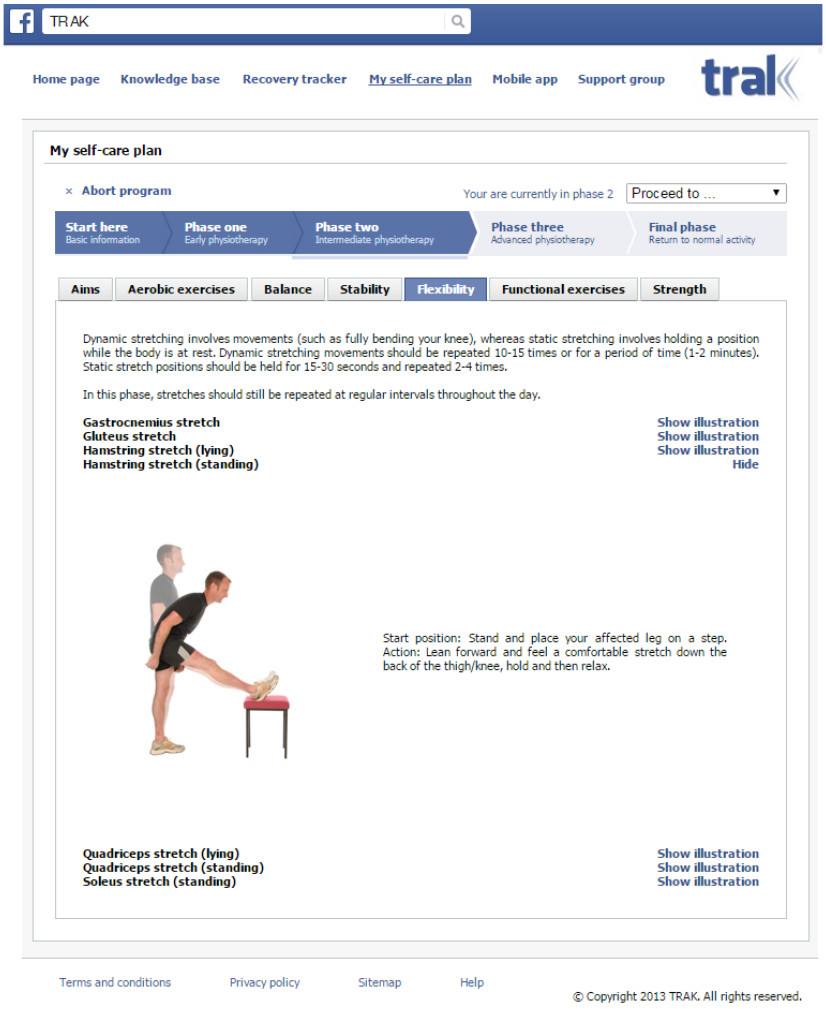


Figure 5. Partially expanded TRAK ontology provides the classification of exercises.

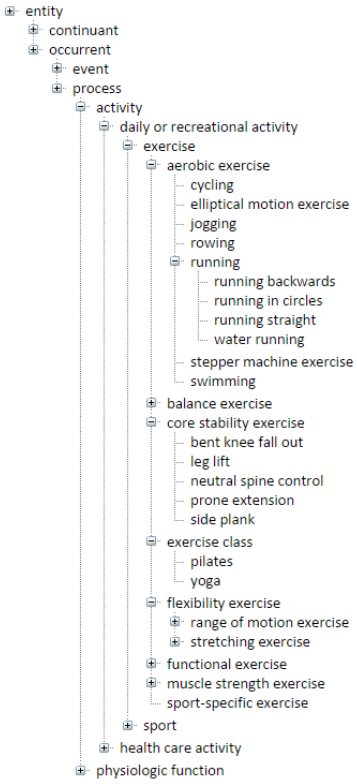
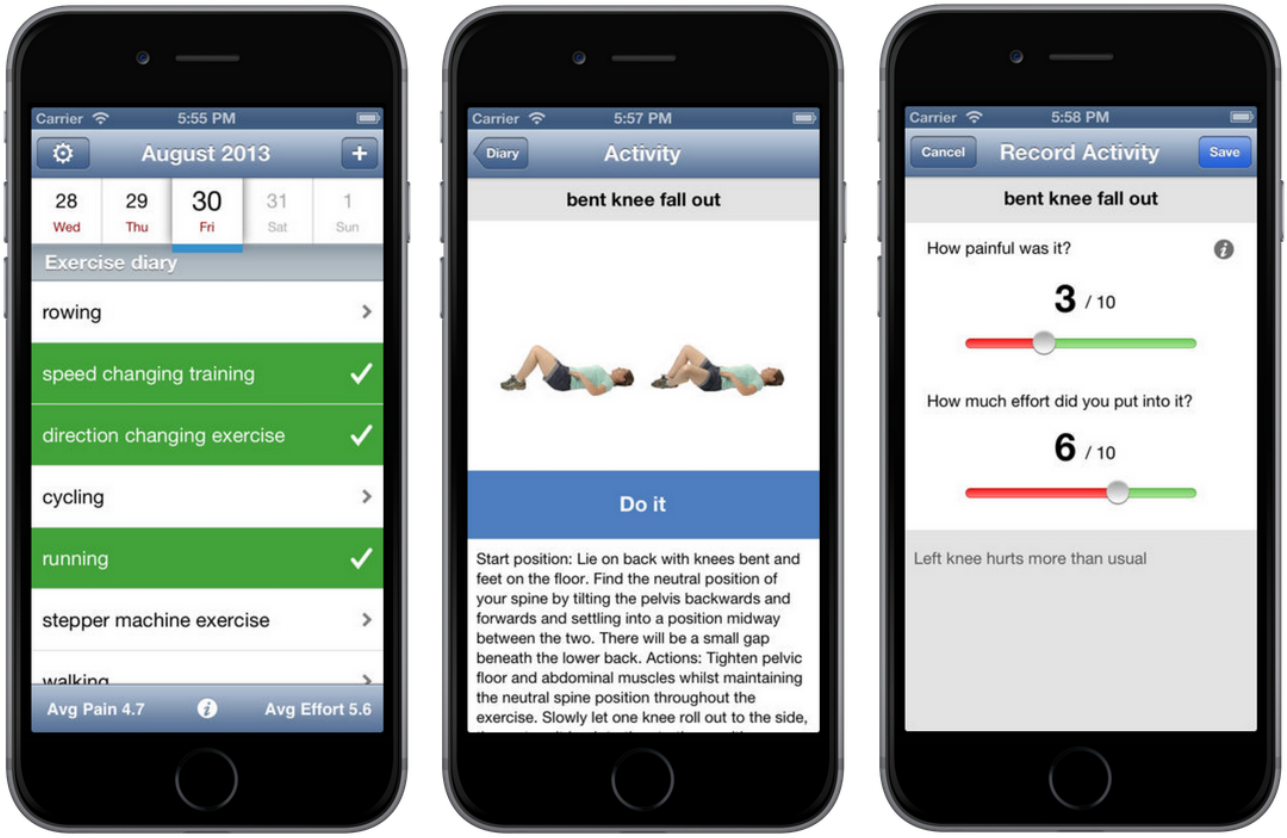


Figure 6. TRAK mobile app allows a patient to keep track of exercises on the go.



Virtual Support Group

The evidence suggests that the capacity of patients to self-manage can be sustained through the perceived social support [67]. In particular, social support proved to be an important external factor influencing how much participants exercise [68]. Therefore, we set up the Support Group to provide a venue for patients to share their experiences. It has been implemented as a Facebook group rather than as a discussion forum in order for the Support Group to be more interactive and familiar to the intended user group. Indeed, Facebook groups provide a platform that seamlessly supports communication, sharing, and interaction within a social group gathered around individuals with common interests [69]. Facebook groups come with intuitive privacy settings to control who can see group's membership and content: secret (only members can find the group and see posts), closed (anyone can find the group and see its members, but only members can see posts), and open (anyone can see the group, its members and their posts). To create a private social space for patients, we opted for a secret group. Because the group cannot be found using Facebook's search function, new members can be added by invitation only. Privacy concerns are addressed through policy and education. In terms and conditions, members are reminded that confidentiality of discussions is expected, but cannot be guaranteed. As with any online communication, anything posted within the group has the potential to become public, thus patients are warned to only post information they are comfortable sharing with others.

Measures

Following the initial app development, its comprehensive testing, and research ethics committee approval, a usability study was conducted to evaluate the usability of the TRAK app suite. There were 3 types of participants that were recruited for the study: software experts, patients, and physiotherapists.

Participants

A total of 44 participants (29 patients and 15 physiotherapists) were recruited through the Physiotherapy Department at the Cardiff and Vale University Health Board. Eligibility criteria were (1) an ongoing knee condition for patients and specialist interest in knee rehabilitation for physiotherapist, (2) aged 18 years or older, as we targeted adult users, and (3) having used a Facebook account at least once a week for more than a month. Potential participants were excluded if they (1) had no Internet access at home, (2) did not speak the English language with native-like proficiency, or (3) had contraindications for physical activity without medical supervision. All participants signed the informed consent document. No incentives were offered for participation in the study.

Prior to approaching patients and physiotherapists, 3 software experts were recruited to provide initial feedback on usability. They were asked to register as users on the app and complete a set of tasks to provide an informed opinion about potential usability issues and suggest possible improvements. They provided written feedback, which was then analyzed to identify the following usability themes: design, feedback, format, instructions, navigation, terminology, and learnability. These themes provided the basis for the development of the usability

questionnaires, which were used to collect both qualitative and quantitative information.

Questionnaires

The questionnaires were divided into 3 parts: (1) Questionnaire 1: computer and Internet usage; (2) Questionnaire 2: task completion; and (3) Questionnaire 3: subjective user preferences.

All 3 questionnaires were accessible online. Patients were recruited during an appointment at the clinic and were instructed to complete Questionnaire 1 at home and use the app as part of their self-care. They completed Questionnaires 2 and 3 at the clinic during the follow-up appointment. Of the 29 patients, 13 did not actively participate in the study. Of the 16 remaining patients, only 1 did not complete Questionnaire 1, but they all completed both app-specific Questionnaires 2 and 3. The responses of all 16 patients were used to provide the summary reported here. Of the 16 patients, 10 were men and 6 were women, whose age ranges were 22.83 ± 4.36 years and 23.60 ± 3.24 years, respectively. As for the physiotherapists, all 15 participants completed all 3 questionnaires. Of the 15 physiotherapists, 11 were women and 4 were men, whose age ranges were 36.45 ± 4.59 years and 40.25 ± 4.57 years, respectively.

It is argued that 5 participants would reveal 80% of usability problems [70,71]. Therefore, a total of 34 (3 + 16 + 15) participants seem sufficient in the context of this usability study. In particular, we have at least five participants from both types of stakeholders: patients and physiotherapists. Their total numbers (16 and 15, respectively) are also evenly balanced, which should provide a similar level of insight into their views on usability.

Results

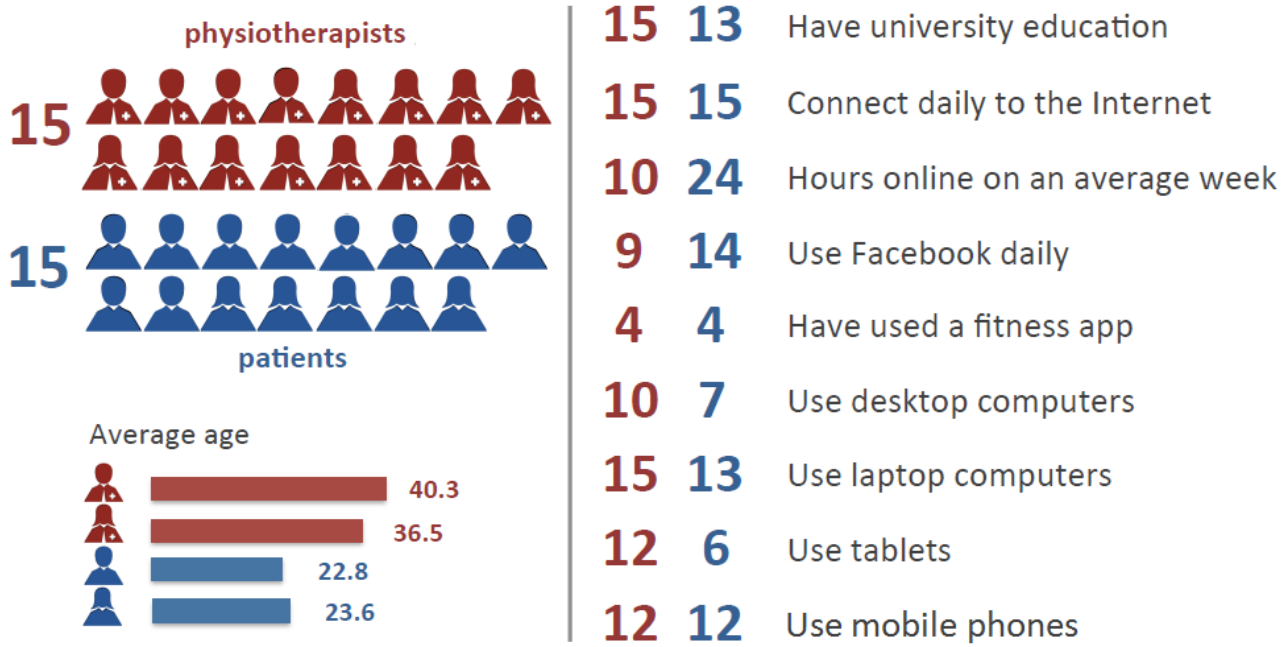
Questionnaire 1: Computer and Internet Usage

Questionnaire 1 consisted of 19 questions whose main purpose was to verify the computer literacy of the participants as part of the inclusion criteria, but also to try and relate the response to task-based questions to different levels of computer literacy. Figure 7 shows a summary of responses to Questionnaire 1.

Of 15 patients who completed Questionnaire 1, all had at least secondary education out of which 13 had a university degree. All of them were accessing the Internet daily (on average 24.2 hours per week) at home mostly via a laptop (87%, 13/15). All 15 patients were mostly using the Internet to send or read email and access an SNS. Of the 4 SNSs (Facebook, Twitter, LinkedIn, and Google Plus), Facebook was the only site regularly used by all participants, followed by Twitter, which was used by 9 of 15 patients. LinkedIn was never used by 13 of the patients, and Google Plus was never used by 12 of the patients. With the exception of 2 patients, all others were likely to use Facebook on a typical day. They used Facebook mostly to communicate with friends and access content on Facebook pages, and somewhat less often to participate in Facebook groups. Almost half of the patients never used Facebook apps prior to this study. Laptops and mobile phones were the most used devices to access Facebook, with laptop being the preferred choice. Most patients (73%, 11/15) have never used an app such as Weight Watchers or iFitness to get in shape. The 4 patients who have used such apps liked their mobile aspect and the ability to use them on-the-go to track exercise performance, but they disliked crashes, frequent updates, large downloads, cost, and some of the apps not being available on Android devices.

Of the 15 physiotherapists, all held a university degree. All of them were accessing the Internet daily (on average 10 hours/week) at both home and work mostly via a laptop (100%, 15/15). All 15 physiotherapists were mostly using the Internet to send or read email and search for information online. Of the 4 SNSs (Facebook, Twitter, LinkedIn, and Google Plus), Facebook was used by 14 physiotherapists, 12 of whom used it on a regular basis (ie, weekly or daily). However, in contrast to patients, only 5 physiotherapists were likely to use Facebook on a typical day. They used Facebook mostly to communicate with friends and access content on Facebook pages, and less often to participate in Facebook groups. A total of 11 physiotherapists never used Facebook apps prior to this study. Laptops, tablets, and mobile phones were the most used devices to access Facebook, with tablets being the preferred choice. Like patients, a majority of physiotherapists (73%, 11/15) have never used an app to get in shape. The 4 physiotherapists who have used such apps liked their ability to demonstrate exercises and track performance, but in addition to frequent updates, large downloads, and cost, they also disliked their effect on battery life as well as some not being user friendly.

Figure 7. A summary of responses about computer and Internet usage.



Questionnaire 2: Task Completion

As part of completing Questionnaire 3, participants were asked to complete a series of prespecified tasks using the app. A total of 15 tasks were defined in order to test the usability of all aspects of the app. The behavior of each user was studied in an individual session. Screencasting software [72] was used to record activities on the computer screen during the task-based sessions. The videos were analyzed to explain any difficulties that lead to incorrect answers to task-based questions.

A total of 16 patients completed Questionnaire 2, most of whom successfully completed the tasks associated with the questions. A total of 10 patients (62%, 10/16) successfully navigated to the Knowledge Base and correctly identified the number of knee conditions described. The remaining 6 patients incorrectly identified the number of knee conditions as 3, which is the number of subheadings under the Knee Conditions tab: Anatomy, Knee Symptoms, and Knee Conditions. This feedback was later used to rename the tab to a more appropriate and less confusing title—“Knee.” A total of 12 patients (75%, 12/16) successfully accessed information via External Links provided in the Knowledge Base. All but 1 patient was able to access the training video on YouTube.

A majority of patients (62%, 10/16) were not able to correctly identify the connections on the Sitemap, either because they were not able to interpret the Sitemap diagram or because they misinterpreted the question and ignored the Sitemap completely. The remaining 6 patients correctly identified that the Mobile App module was only directly linked to My Self-Care Plan. To rectify this, we added a reference to Sitemap to the Home page and explained that its purpose is to illustrate the structure of the app through the use of pages, tabs, and link. Further, all patients were able to identify the currently selected module, report their latest KOOS, and check when their next pedometer reading was due for submission.

A total of 10 patients (62%, 10/16) successfully navigated to the iTunes page from which the mobile app was available for download. The remaining 6 patients failed to scroll down to be able to see a link to iTunes. For this reason, we moved the link higher up, as it was not visible on the laptop screen used during the task-based sessions. Most patients (75%, 12/16) did not own an iOS-compatible device, and, therefore, were not able to perform the tasks using the mobile app. In response, we implemented an Android version of the mobile app.

A majority of patients (81%, 13/16) successfully identified the number of exercises described in Phase 3 of My Self-Care Plan. The remaining 3 patients counted the number of strength exercises in Phase 1 instead. Most patients (88%, 14/16) were able to correctly interpret the description of a specific exercise, but 2 failed to correctly describe it as they seemed to ignore the explicit textual description, which stated "Start position: Stand upright. Action: Lift the unaffected leg...." For example, one patient stated the start position as "stand on left leg" by literally interpreting the image provided. The other one misinterpreted the description as "affected knee off floor...."

All but 1 patient successfully identified the privacy settings in the Support Group. However, most patients (81%, 13/16) were not aware of the types of personal information stored by the app even though it is clearly specified in the privacy policy. To direct users to this information, we added a reference to the Privacy Policy on the Home page. We also added an explanation on how to delete an account, together with the associated information.

A total of 15 physiotherapists completed Questionnaire 2, most of whom successfully completed the tasks associated with the questions. Only 1 physiotherapist incorrectly identified the number of knee conditions as 3, which was again attributed to an ambiguity in naming the tab under which the subheading “Knee Conditions” is located. As indicated earlier, the tab in question was subsequently renamed to better reflect its content.

A total of 11 physiotherapists (73%, 11/15) successfully accessed information via External Links provided in the Knowledge Base. A total of 8 physiotherapists were able to access the training video on YouTube, but the remaining 7 have not attempted to look for the training video.

A slight majority of physiotherapists (53%, 8/15) were able to correctly identify the connections on the Sitemap; 4 other physiotherapists misunderstood the questions, but 3 correctly provided the total number of connections. The remaining 3 physiotherapists misinterpreted the question and ignored the Sitemap completely, thus not providing a correct response. We explained the corrective action taken previously in response to patients' feedback. Like patients, all physiotherapists were able to identify the currently selected module, report their latest KOOS, and check when their next pedometer reading was due for submission.

All but 1 physiotherapist successfully navigated to the iTunes page from which the mobile app was available for download. As indicated earlier, we moved the link higher up to make it visible without the need to scroll down. While most patients (75%, 12/16) did not own an iOS-compatible device, a majority of physiotherapists (67%, 10/15) did own one, which allowed us to get more insight into the usability of the mobile app. All of them were able to select an exercise and record information such as pain level. Only 1 physiotherapist did not manage to find previously recorded information.

A majority of physiotherapists (93%, 14/15) successfully identified the number of exercises described in Phase 3 of My Self-Care Plan. There was 1 physiotherapist who incorrectly entered the number of exercises as 6, but the analysis of the video showed them pointing at the 5 exercises provided, so we assume that this was a typographical error. Partly relying on their specialist knowledge, all physiotherapists were able to correctly interpret the description of a specific exercise.

All but 1 physiotherapist successfully identified the privacy settings in the Support Group. Still, like patients, the vast majority of 12 physiotherapists were not aware of what types of personal information were stored in the app. As indicated before, we made an explicit reference to the Privacy Policy on the Home page with an explanation on how to delete information from the app.

Questionnaire 3: Subjective User Preferences

Questionnaire 3 was designed to highlight subjective user preferences about the app including its general usability, user perception, and appropriateness in the context of exercise-based rehabilitation. The part about general usability was based on the System Usability Scale (SUS), a questionnaire for assessing the perceived usability of interactive systems [73]. It consists of 10 questions based on a 5-point Likert scale (1=strongly disagree, 5=strongly agree). In comparison to other commonly used questionnaires, it was shown to be the simplest and most reliable in determining website usability [74]. Not surprisingly, it is the most used questionnaire for measuring perception of usability. The overall SUS score is calculated on a scale from 0 to 100. The widespread usage of the SUS questionnaire allows the usability of a system to be benchmarked against others. Based on the average SUS score, any score above 68 is considered above average. In our case, the SUS score calculated from patients' responses was 78, whereas that of physiotherapists was 75, which belong to a percentile range of 80-84% and 70-79%, respectively. In other words, our app has higher perceived usability than 70% of systems. This rank can be interpreted as “Grade B” on a scale from A to F [75].

Figure 8 shows a summary of responses about user perception. Only 1 patient out of 16 found the app unnecessarily complex, cumbersome, inconsistent, difficult to learn, not easy to use, and not well integrated. None thought they would need technical support to use the app. Similarly, a single physiotherapist out of 15 found the app unnecessarily complex and not easy to use. There were 3 other physiotherapists that thought that they would need the support of a technical person to be able to use the app. They also did not feel confident using the app. By contrast, various functions in the app were found to be well integrated with no inconsistencies. All physiotherapists agreed that most people would learn to use the app very quickly. It seems that the physiotherapists experienced more difficulties in using the app than the patients, which was reflected in a slightly lower SUS score.

In addition to SUS-based questions on general usability, we formulated 8 additional diagnostic questions. All patients agreed that the information provided was credible, useful, and easy to understand. The physiotherapists agreed with patients' views that the information provided was credible, useful, and easy to understand. However, 1 physiotherapist disliked the layout of the app, and another disliked the presentation of the app.

Finally, 14 questions were asked in relation to the appropriateness of the app in the context of exercise-based rehabilitation. The patients unanimously agreed that exercise descriptions were sufficiently clear and that the images matched the description well and made it easier to follow the instructions. However, 12 of 16 patients (75%) said that they would prefer videos to images. Most patients (69%, 11/16) felt encouraged to exercise more. With the exception of only 2 patients, all others felt able to progress the exercises using the app and that it improved the self-management of their knee condition. A slight majority of patients (56%, 9/16) believed that using the app facilitated the recovery from knee condition. A total of 14 patients (88%, 14/16) would recommend the app to a friend, colleague, or family member who suffered from a knee condition. Only 1 patient was concerned about the privacy or security regarding the app's use.

When asked about an optimal number of exercises that should be available in the app, most physiotherapists explicitly stated that the current content was appropriate in terms of quantity and variety. With the exception of a single physiotherapist, the rest unanimously agreed that exercise descriptions were sufficiently clear, and that the images matched the description well and made it easier to follow the instructions. Similarly to patients, 11/15 physiotherapists (73%) said that they would prefer videos to images. In line with the patients' views, the majority of physiotherapists (67%, 10/15) believed that patients would be encouraged to exercise more. With the exception of 1 physiotherapist, the professional opinion of the remaining 12

physiotherapists (80%, 12/15) was that the information provided was sufficient to be able to progress the exercises. More physiotherapists (11/15) than patients (9/16) believed that using the app could facilitate the recovery from a knee condition. Patients were seen 2 weeks following the recruitment, which is a relatively short period for patients to observe a significant recovery, but obviously the prevalent professional opinion based on clinical practice was that such recovery would be noticeable over time. The same subset of 11 physiotherapists agreed that using the app could improve the self-management of a knee condition. All 15 physiotherapists would recommend the app to someone who suffered from a knee condition. Whereas only 1 patient was concerned about the privacy or security regarding the app use, 4 physiotherapists expressed such concerns.

We analyzed patients' opinions on whether the app could support face-to-face appointments with a physiotherapist and extracted 6 main themes given in Table 1. Likewise we also analyzed physiotherapists' opinions (Table 2), which revealed the overall agreement on the given issues from 2 different perspectives.

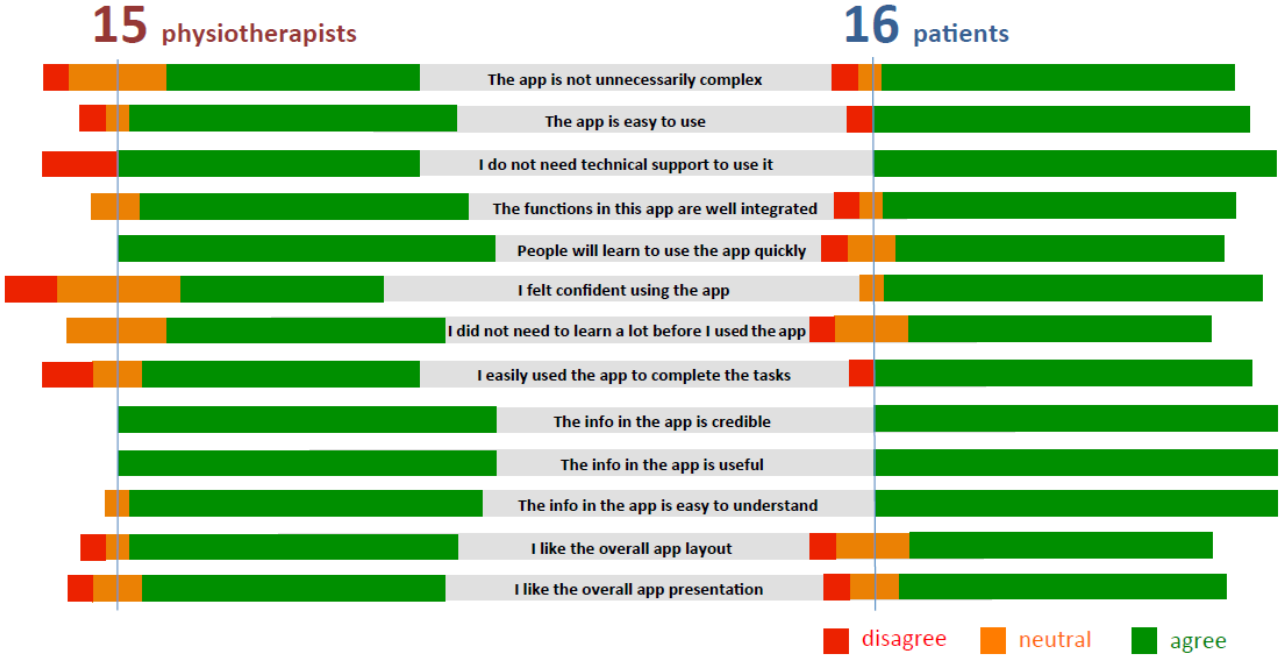
Table 1. A sample of patients' opinions on the extent to what the app could support face-to-face appointments.

Theme	Example
Facilitate communication	It could form a good trigger for talking points or examples; for example, I tried the third knee stability exercise and really struggled with it.
Improve understanding	If unsure of any exercise, I can easily access the app to clear any confusion.
Provide information	It gives extra exercises I can use on top of what my physiotherapist has given me.
Support self-management	Providing a head start to complete the tedious exercise on your own.
Enable progression	It would be a good reminder of the exercises explained by your physiotherapist and how to do them, and encourage you how to progress them if they become too easy.
Recall information	It can be a good reminder of the exercises the patient is supposed to do. I have often come away from a physiotherapy appointment with so many exercises swimming round in my brain that I get them all a bit mixed up.

Table 2. A sample of physiotherapists' opinions on the extent to what the app could support face-to-face appointments.

Theme	Example
Facilitate communication	This app would streamline discussions to relevant areas, saving the clinician's (and patient's) time.
Improve understanding	Improve ability to check exercises against a clear picture and description.
Provide information	The quality and accessibility of information are much better than any that we currently provide at face-to-face rehabilitation sessions.
Support self-management	I think this is a great resource, which can back up and add to the information provided by therapists and allow patients to be much more in control of their own rehabilitation strategies.
Enable progression	Physiotherapist can also highlight exercises to progress onto knowing the patient will have a clear instruction at the appropriate time.
Recall information	Good as a reminder of exercise technique and to stop the need to draw stick men exercises.

Figure 8. A summary of responses about user perception.



Face-to-Face Appointments and the App

Further, we analyzed the patients' opinions on whether the app could replace face-to-face appointments with a physiotherapist. The overwhelming majority of patients (n=10) felt strongly that while the app would be a useful aid to their rehabilitation, it could by no means substitute face-to-face contact. The main reasons they provided were related to the reassurance associated with human contact as well as the potential to provide personalized care. However, 3 patients believed that, while the app could not completely replace face-to-face appointments, it could certainly reduce the number of such appointment. There were 2 patients who found this aspect particularly useful, quoting logistic reasons such as time constraints and transport links, which at times make it difficult for them to attend appointments. This implies that the app would be of high value to patients with restricted access to health care premises, for example, because of impaired mobility, remote locations, and/or nonflexible working conditions.

Not surprisingly, the physiotherapists correctly predicted that patients would not be keen on fully replacing face-to-face appointments. However, they themselves were more open to the idea of reducing the number of face-to-face appointments (10 of 15 physiotherapists), typically suggesting alternative forms of contact such as telephone or Skype conversations, but also embedding a direct contact into the app itself. In contrast to patients who specified reassurance as the main reason for insisting on face-to-face appointments, physiotherapists named monitoring and progression as their professional reasons.

Finally, when asked about any other comments, 5 patients provided no additional comments, 8 patients were extremely positive about the app (Table 3), and 3 left very specific comments about particular aspects of the app (eg, number of clicks, amount of information). A total of 11 physiotherapists shared the overwhelmingly positive reaction to the app (Table 4). There were 3 physiotherapists who left specific comments about the ways of improving the app (eg, adding contact option, measuring adherence).

Table 3. A sample of patients' comments.

Theme	Example
Uptake	The TRAK app is well set out and it's easy to access different areas. The information provided is thorough and clears any confusion about any knee problems. I will indefinitely be using it as of today!
Progression	Great app in general and very helpful, exactly what's needed to securely help patients along the road to recovery.
Content	I thought the app was exceptional, really interactive and very clever. My only comments would be for the information surrounding the injury/knee joint (medical bit) could be slightly shorter worded and perhaps be written in bullet-point form.
Technology	I rarely used the Facebook app, was barely aware Facebook did apps. If the full (Facebook version) of this app was available for iPhone with the instructions, plan etc, I would have used it a lot more and found it far more useful.

Table 4. A sample of physiotherapists' comments.

Theme	Example
Uptake	May be useful to add a goal setting function/monitor progress with specific functional/fitness goals. Overall, think it is an excellent idea and would definitely consider using it with patients, clearly a lot of work gone into developing the app/information & exercises. I look forward to becoming more familiar with it and using it more.
Progression	Looks really good. Would be great to build a database to see how a cohort of knee conditions progress through their rehab and return to function/work/sport. Physiotherapists could use this information to spot those failing to progress earlier, and help inform patients more accurately about their prognosis/timescales.
Content	Good. I think videos of patients doing the exercises in a good or bad way would be even greater.
Technology	Great idea for younger/tech savvy patients who are self motivated. Would there be a pop up reminder regarding follow up appointments in physiotherapy? Might be useful if appropriate.

App Feedback

In conclusion, the results of the usability study informed the subsequent improvements of the app by considering all aspects that have an impact on user experience. This section outlined a set of measures taken in response to issues raised in user feedback. Minor changes included renaming and rearranging the content of the app to improve navigation, content, and facilities designed to help users' learning experience.

A major change was the addition of an Android app to the app suite. This issue was first identified in patients' response to Questionnaire 1 (computer and Internet usage), when patients who had prior experience with using fitness apps raised an issue of some of these apps not being available on Android devices. The need for an Android version of the mobile app became obvious when the responses in Questionnaire 2 (task completion) highlighted that the majority of patients (75%, 12/16) did not own an iOS-compatible device. Indeed, these results are in line with data reported by the International Data Corporation [76], a global provider of market intelligence for the information technology, telecommunications, and consumer technology markets. According to their Worldwide Quarterly Mobile Phone Tracker, Android dominates the mobile phone operating system market share, accounting for nearly 85% in comparison to iOS, which accounts for only 12%. Android's market share has been increasing steadily over the last 4 years from 36%, 70%, 80%, to 85% during the period from 2011 to 2014. Obviously, Android is by far the most popular operating system on mobile phones. Therefore, ignoring this platform would alienate a vast majority of potential users of the TRAK app suite. The Android version of the mobile app was implemented following the conclusions of the usability study. We incorporated information about it into the Mobile App module of the Facebook app. The 2 mobile apps are equivalent in their functionality and both can be used as standalone products.

Discussion

Limitations of the Study

This paper described a novel Web-based app for delivering exercise to patients undergoing knee rehabilitation and provided preliminary usability findings. A limitation of the usability study was the discrepancy between the age range (19-31 years) of the patient sample and the fact that the risk of developing osteoarthritis increases from the late 40s [2]. Potential usability issues related to the app design in the context of the physical abilities and the visual capacity of older users can be resolved relatively easily. A major concern, however, is that of digital literacy across the age groups. Not surprisingly, Internet usage was found to fall with age across Europe, but the increasing development of useable and accessible products such as mobile phones and mobile apps is expected to reduce the challenge of digital literacy [77]. In Europe, the Riga Declaration 2006 [78] established specific targets in relation to aging and information and communication technologies, one of which is to halve the gap in average Internet use between older people and the EU population. In this context, the usability results can be generalized beyond the age range of the patient sample used in the study.

The usability study uncovered various areas of possible improvements. Both patients and physiotherapists suggested that videos should replace images as illustrations of exercises. We already produced over 20 exercise videos. From a developer's perspective, replacing images with videos would constitute only a minor change to the system. Although quoting different reasons, all stakeholders expressed a wish for the app to close the feedback loop between patients and physiotherapists by allowing them to exchange information and communicate directly as part of a shared decision-making process. Indeed, support for patient-doctor interactions was identified during needs assessment. Incorporating a Web chat function [79] into the app would allow a patient to chat with a physiotherapist in real time. In technical terms, it would simply require a developer to reuse a piece of ready-made code. The reason we did not implement this functionality was due to human resource constraints. While it would be worth investing in this particular aspect of a Web-based intervention, the limitations on human resources are likely to persist. A fully automated question-answering system (QAS) or at least a list of FAQs would provide a compromise solution to this problem. As part of piloting the Web intervention within the NHS, plans are already in place to collect FAQs and incorporate them into the TRAK app suite. In combination with the TRAK ontology, the collected questions and answers will provide a basis for further research into developing a QAS.

Another possibility to improve the function of the support group would be to involve a health care professional as a moderator. Namely, such groups provide a communication space, but not a self-sustaining conversation. A moderator strategy is required to support community development. There are 2 ways of

moderation that are essential for health promotion interventions: (1) professional supervision to maintain a safe space for discussion and information quality, and (2) a more engaged presence to improve activity and timely response to user requests [80].

Future work will support the professional needs of physiotherapists by allowing them to specify exercise prescription within the app as part of personalizing its exercise program according to individual patients' circumstances. The TRAK ontology will drive the exercise selection process. The ontology will also facilitate tighter integration of the individual apps within the TRAK suite. In particular, we want to exemplify the correlation between exercise adherence (currently monitored by the mobile app) and recovery progression (currently quantified by the KOOS within the Facebook app). In addition to providing feedback and motivation to patients, such information would allow physiotherapists to monitor patients' recovery and exercise progression. On a large scale, these data can support epidemiological studies to identify the most effective treatment components, so that new interventions can be developed.

The assessment of long-term impact of a Web-based intervention on knee rehabilitation was outside the scope of this study. Nevertheless, our work laid out the foundation for further translational research based on a randomized control trial. We recently acquired funding to put the TRAK intervention into practice within the Cardiff and Vale University Health Board and gather evidence about how such innovation improves quality of health care. This will provide an opportunity to explore the link between face-to-face physiotherapy interaction and the use of the app in light of the finding that better outcomes in Web-based interventions were identified when there were multiple contacts with participants and when the time to follow-up was short [14].

Conclusions

The aim of this study was to assess the usability and acceptability of a Web-based intervention in knee patients and physiotherapists who deliver knee rehabilitation. We developed TRAK, a Web-based app suite, to support self-management of knee conditions. Its content is based on the TRAK ontology, which includes rehabilitation concepts and treatment modalities that are part of standard care for the rehabilitation of knee conditions based on expert clinical opinion and published research evidence [8]. The usability study suggested unanimous acceptability by both types of stakeholders. Both patients and physiotherapists agreed that the given Web-based approach would facilitate communication, provide information, help recall information, improve understanding, enable exercise progression, and support self-management in general. The Web app was found to be easy to use and user satisfaction was very high. These results suggest that a Web-based intervention is feasible and acceptable in supporting self-management of knee conditions.

Acknowledgments

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Authors' Contributions

IS and KB conceived the overall study and drafted the manuscript. TP originally designed and implemented the Facebook app. IS and KB provided user feedback for the first round of modifications implemented by TP. All subsequent modifications were implemented by IS. KB, RvD, and CW provided the content of the app. DP and AP designed the mobile app, which was then implemented by DP for iOS. SG implemented the Android version of the mobile app. IS, KB, and AD designed the usability study and coordinated usability testing. With the exception of IS and KB, all other coauthors are listed in alphabetical order.

Conflicts of Interest

None declared.

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Abbreviations

FAQ: frequently asked questions
KOOS: Knee injury and Osteoarthritis Outcome Score
NHS: National Health Service
QAS: question-answering system
SNS: social networking site
SUS: System Usability Scale
TRAK: Taxonomy for RehAbilitation of Knee conditions

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Original Paper

Prescription Tablets in the Digital Age: A Cross-Sectional Study Exploring Patient and Physician Attitudes Toward the Use of Tablets for Clinic-Based Personalized Health Care Information Exchange

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Abstract

Background: To reduce the cost of health care while increasing efficiency and quality, health systems are seeking innovative means to engage and empower patients. Improved use of information technology and electronic health record (EHR) infrastructure is essential, and required for "meaningful use" as mandated by the federal government. Providing personalized health information using tablets at the point of care could enhance the clinical experience and enable efficient collection of patient reported outcome measures to guide clinical decision making.

Objective: The aim of this study is to explore patient and provider attitudes and interest in a proposed clinic-based tablet system for personal health information exchange. To provide a context to understand patients' use of tablets during their clinic visit, we also examine patients' current activities and time spent in the waiting room, and their use of health information resources.

Methods: Surveys were administered to 84 patients in the waiting room of a community health center affiliated with Massachusetts General Hospital (MGH) in Boston, MA. This survey included a vignette and illustration describing a proposed tablet-based system in which the patient, upon sign in at the clinic, receives a tablet loaded with personalized information tailored to their specific medical conditions and preferences. Patients were queried about their interest in such a system in comparison to traditional forms of patient education as well as their current health information seeking behaviors and activities and time spent in the waiting room. Interviews with five MGH-affiliated health care providers were conducted to assess their opinions regarding the proposed tablet system.

Results: The majority (>60%) of patients were "very" or "extremely" interested in the proposed tablet system and thought it would improve their knowledge about their medical condition (60%), assist them in making healthy choices (57%), and help them to feel more comfortable talking with their provider (55%). Patients thought the system would be more motivating, informative, and engaging than traditional printed health education materials. The tablet system was not considered more effective than face-to-face interaction with providers, though 44% thought it would improve their relationship with their physician. Overall, 91% of respondents were willing to learn how to use a tablet and 75% reported being "very" or "extremely" confident they could

use one. Four of the five providers believed that the proposed tablet system would improve clinical workflow and patient education. Patients and providers were concerned about privacy and security of data collected using the tablets.

Conclusions: Both patients and providers were highly amenable to integrating tablets into the clinical experience, and tablets may be useful in improving patients' health knowledge, the collection of patient reported outcome measures, and improved patient-provider communication. Further research into operationalizing such systems and their validation is necessary before integration into standard clinical practice.

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KEYWORDS

electronic health records; tablets; health education; self-management; primary care; physician patient relationship; digital health; iPad; patient-reported outcome measures

Introduction

Total health care expenditure in the United States is expected to reach US $4.4 trillion dollars by 2018, comprising over a fifth of total gross domestic product (GDP) [1]. Greater than three fourths of this expenditure will be related to chronic medical conditions, which can either be improved or prevented given the appropriate lifestyle modifications [2]. These staggering statistics highlight the importance of promoting behavior change, healthy lifestyles, and chronic disease self-management—not only to help control the rising cost of health care, but also to actively engage patients in their own wellness, reduce morbidity, and improve patient satisfaction and quality of life. Unfortunately, many teachable moments for patients are missed due to increasing educational tasks of health care providers and time constraints [3].

For example, it is well-known that regular physical activity can improve blood pressure, blood glucose control, and quality of life, while lowering harmful cholesterol levels and decreasing cardiovascular events and mortality [4-7]. Although providers understand the importance of their role in engaging patients to participate in regular exercise, they report that the lack of time and competing demands are significant barriers in providing this service and less than one third of primary care visits include any exercise or lifestyle counseling at all [8,9]. In an era when medical trainees have been found to only spend 12% of their time (as little as 8 minutes per patient) examining and talking to patients and more than 40% behind a computer, it is of utmost importance to find innovative solutions to provide patients and providers with a more meaningful interaction [10]. Additionally, with the advancement of the Meaningful Use criteria set forth by the federal government, electronic health records (EHRs) must adapt to provide additional control for patient's data and demonstrate an improvement in patient outcomes [11].

As described by Sinsky et al [12], we have come to a time in the evolution of EHRs that the power of these technologies to support the needs of primary care providers and patients must be better utilized. New electronic information tools should not only add value to the interaction with a patient-centric design, but must also allow providers the opportunity to improve efficiency and align with the goals of the patient-physician relationship [12]. Primary care is at a turning point in history and recognizing the need to improve these aspects with new technologies will help steer future physicians to this rewarding and much needed profession [12]. One potential method of optimizing workflow, decreasing the burden on providers, and increasing the potency of an office-based visit is by leveraging the use of digital technology, such as tablets, to increase patient knowledge and self-management [3,13-15]. This method also makes more efficient use of the limited time that a provider has with the individual patient in that tablets can be used to collect and integrate pertinent patient data prior to the face-to-face clinical encounter, allowing providers to focus on the interaction with the patient and higher level analysis rather than data entry [16-19].

Since the advent of the extremely popular iPad in 2009, the health care community has widely adopted its use as a reference in clinical practice [20,21]. Touchscreen tablets, however, are also increasingly being employed to provide education to patients when they come into the clinic, rather than just a reference tool for providers [20,22]. Organizations such as the Mayo Clinic and the Cleveland Clinic have developed unique patient education applications on various medical conditions and a quick search of the Apple App Store reveals hundreds of such applications [23,24].

Questions arise as to the ubiquitous use of tablets in patient education and engagement for a variety of reasons. While it has been shown that patients who were older, had lower annual household income, and lower educational attainment had more difficulty using advanced communication technologies, a vast majority (94%) of patients across all socioeconomic status backgrounds rated tablet devices as easy to use [25]. Further, by using audio-visual digital media, even low-literacy patients and those recovering from major surgery could be educated about their medical conditions effectively [26,27].

Although many systems have been devised to apply information technology to engage and educate patients, none have achieved widespread use [13]. In the face of a rapidly evolving health information technology (HIT) infrastructure, it is important to create standardized systems that are easy to use and truly afford patients more access to and control over their health data [28]. Prior studies examining the clinical effects of computer-based education and self-management have shown modest benefits on a range of conditions: blood sugar control in diabetes, weight loss after Roux-en-Y gastric bypass surgery, and adherence to immunosuppressive medications after lung transplantation [29-31]. Utilizing computerized educational tools such as kiosks has also been shown to increase patient knowledge on a range of conditions, including the importance of HIV screening,

appropriate antibiotic use for upper respiratory tract infections, and adequate asthma care [32-34]. Although a tablet-based patient education system could have many potential benefits similar to the kiosk model, the form factor lends itself more to rapid adoption and scalability. Currently, there is a paucity of evidence regarding patient and provider perceptions of the benefits and barriers of using tablets in the primary care setting.

With this formative study, we sought to explore both patient and provider interest in using tablets for personalized health information exchange in the primary care setting. We hypothesized that this technology could be a powerful catalyst in transforming the health care experience, creating a platform for just-in-time patient education, intelligent intake exams, and collection of patient-reported outcome measures (PROMs). This strategy also takes advantage of the psychology of physically coming to the clinic where patients give their time and attention in hope of receiving personal health care and advice. We also sought to understand patients' current activities and the amount of time spent in the waiting room, and their health information use, needs, and preferences. The results of this formative research will help guide the design of future clinic-based HIT systems to increase patient knowledge, engagement, and satisfaction while improving provider efficiency and outreach.

Methods

Recruitment

The study was developed at Connected Health Innovation (CHI) in Boston, Massachusetts, and approved by the Institutional Review Board of Partners HealthCare and Massachusetts General Hospital (MGH). A research intern administered the survey portion of the study by approaching patients in the waiting room of primary care physicians at an MGH-affiliated community health center in the greater Boston area. This clinic was selected because it is located in an ethnically and socioeconomically diverse community that is relatively representative of patients in the Boston metropolitan area. Eligibility criteria included age 18-75 years and patient status at the clinic. Due to the formative nature of this study, the survey was available only in English and participation was restricted to patients who could speak English. Participation was voluntary and a small remuneration of US $5.00 cash was provided to patients in appreciation for their time. Of the 194 people approached, 115 (59.3%) agreed to participate and of these, 28 (24%) were excluded for not meeting eligibility criteria. The most common reasons for exclusion were age and inability to speak English (n=24). Of the 87 that met the inclusion criteria, 84 completed and returned the survey.

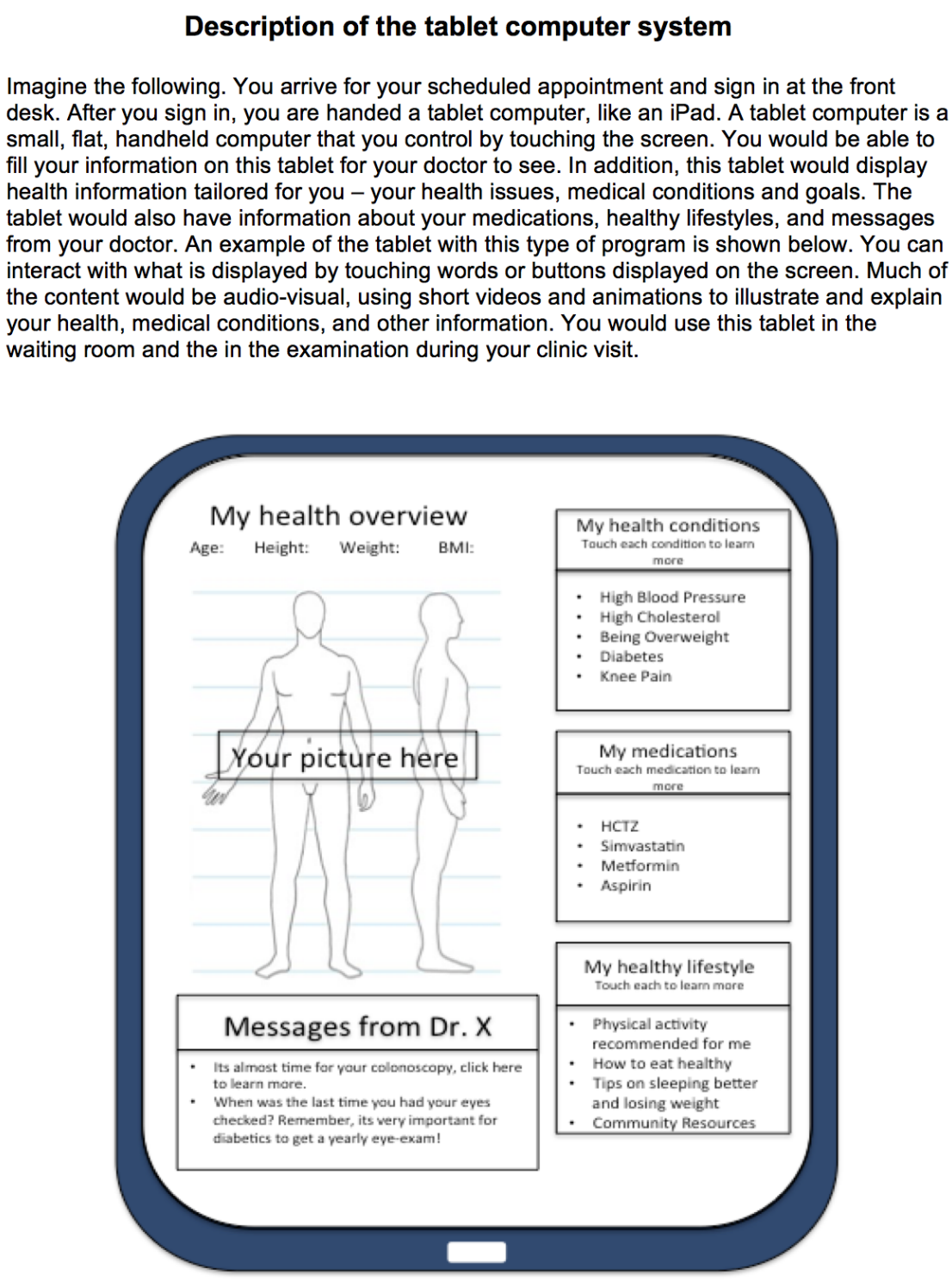
Our second goal was to gather formative data to highlight key points, both positive and negative, that would be of concern to providers in the creation and implementation of a tablet-based system. Five MGH-affiliated providers were recruited using a snowball sampling method in which each provider interviewed recommended another provider to contact. Five providers were contacted via email and all 5 consented to participate in a semistructured phone interview. Providers did not receive remuneration for their participation.

Data Collection

A 16-page paper survey containing 46 questions was administered to patients and took about 15 minutes for patients to complete (see Multimedia Appendix 1). National surveys were used as the source of questions on technology and Internet use (ie, Pew Research Center) and health information sources, patient-provider communication, health care utilization, and sociodemographics (ie, National Cancer Institute's Health Information National Trends Survey). The survey included a description and illustration of the user interface of the proposed tablet system (Figure 1) and a series of questions regarding patients' interest in using this system, types of information they would like to receive, privacy and usability concerns, and attitudes regarding the impact of the system on their health care. Questions about the proposed tablet system and time spent in the waiting room were created by the researchers based on their prior experience studying connected health adoption and use.

A semistructured interview script was created by the research staff to guide the phone interview with providers. The interview covered the practice type and patient characteristics, time and methods used to counsel and educate patients, and perceived efficacy of current education, and patient-provider interactions to improve knowledge and promote healthier behaviors (see Multimedia Appendix 2). Following a description of the proposed tablet system, providers were asked to give their thoughts regarding the proposed system, including general positive or negative reactions, how such a system might be used in the clinic (ie, education, patient-provider communication, decision making, tracking symptoms or patient reported outcomes), potential efficacy, and any concerns on disadvantages or difficulties that might be encountered. Interviews were conducted by a research intern, took about 15 minutes, and an audiorecording was made for transcription and summarizing provider responses.

Figure 1. Diagram and description of tablet-based health information exchange system presented to patients.



Statistical Analysis

Descriptive statistics were generated from the patient surveys using Excel (Microsoft, Redmond, WA, USA) and Stata version 13 (StataCorp, College Station, TX, USA). Transcripts of the interviews with providers were reviewed by the research intern and a research scientist at the Connected Health Innovation (CHI) to summarize providers' responses to questions and general comments.

Results

Patient Characteristics

Characteristics of the patient sample are presented in Table 1. The mean age was 43 years. Thirty-nine percent were male and 72% (60/83) described their race as white, with the largest single minority being Hispanic. A little over half of the sample (57.5%, 46/80) had completed one or more years of college. Most patients rated their health as "good" (37.5%, 30/80) or "very good" (32.5%, 26/80).

Table 1. Patient sample characteristics.

Variable		Participants[a]
Age, mean (SD)		43.05 (13.29)
Sex, n (%)		
	Male	31/80 (38.75)
	Female	49/80 (61.25)
Race/ethnicity, n (%)		
	White	60/83 (72.29)
	Hispanic	5/83 (6.02)
	Black	4/83 (4.82)
	Asian	4/83 (4.82)
	Other	10/83 (11.05)
Marital status, n (%)		
	Married or living with partner	45/80 (56.25)
	Divorced, separated, or widowed	11/80 (13.75)
	Single, never married	24/80 (30.00)
Education level, n (%)		
	1st-11th grade	6/80 (7.50)
	12th grade, completed high school, or GED	28/80 (35.00)
	1 to 3 years of college	25/80 (31.25)
	4 or more years of college	21/80 (26.25)
Has a regular health care provider, n (%)		52/81 (64.20)
Number of times saw physician during past year, mean (SD)		3.15 (1.91)
Self-rated health status, n (%)		
	Poor	3/80 (3.75)
	Fair	12/80 (15.00)
	Good	30/80 (37.50)
	Very good	26/80 (32.50)
	Excellent	9/80 (11.25)

[a]Due to missing data (no response to some of the questions on the survey), there is incomplete data on some questions. Thus, only 80 people filled out the question on sex, 83 completed the race question, etc.

Twenty-four percent (17/70) of patients already own a tablet, 79% (61/77) own a laptop or notebook, and 74% (59/80) own a desktop computer. Seventy percent own a smartphone, 14% (11/78) a "feature" phone (a mobile phone that lacks smartphone features), and 65% (50/77) have a wired telephone. An overwhelming majority (91%, 73/80) of patients said they were willing to learn to use a tablet and 76% (63/83) were "very" or "extremely" confident they could use a tablet if they had to. Only 2% (2/83) said they were "not at all confident" they could use a tablet.

Time Spent in the Waiting Room

The time patients spent waiting to see their doctor during their last visit varied widely with 3 reporting wait times of 90 minutes or longer. The mean wait time was 28.5 minutes with a median of 20 minutes. Taking into account these factors, an estimate of typical wait times is between 20 and 25 minutes.

Fifty-six percent of patients said that the time they spent waiting in the doctor's office was time *not* well spent. We asked patients to choose from a list of 13 activities all of the things they normally do while waiting to see their doctor. The most common

activities were reading the newspaper (63%, 53/84) or sitting quietly (43%, 36/84). A substantial percentage of patients used their personal devices to send or receive text messages (short message service, SMS) (29%, 24/84), browse the Internet (23%, 19/84), or talk on the phone (21%, 18/84). Patients did report spending some time on health-related activities while waiting—24% (20/84) spent some time reading handouts or pamphlets on health topics.

Health Information Seeking, Sharing Information With Physicians, and Information Needs

Eighty-nine percent (74/83) of patients said they had at one time looked for information about health or medical topics. When these patients last looked for health information, the overwhelming majority turned first to the Internet (83%, 52/63), with doctors and other health care providers being placed a distant second (8%, 5/63). During their most recent search, patients were most often seeking information for themselves (56%, 43/77) or for both themselves and someone else (29%, 22/77). Only 13% (10/77) of patients were seeking health information for someone else. Based on patients' experiences during their most recent search for health information, 53% (40/75) were "concerned about the quality of the information" they found. A substantial proportion of patients reported that finding the health information they needed took "a lot of effort" (30%, 23/77) and was "hard to understand" (24%, 18/75).

Although doctors and other health care providers placed a distant second to the Internet as a source of health information during patients' last search, the gap narrowed when patients were asked where they would turn first if they had a strong need to get health or medical information. Fifty-seven percent (44/77) said they would use the Internet and 29% (22/77) said they would turn first to their doctor or health care provider. About 39% (31/80) of patients said they talked with their doctor or health care provider in the last 12 months about the information they found online. However, provider interest in hearing about this information was mixed. Patients said that 40% (17/43) of providers were "not interested at all" or "a little interested", and 60% (26/43) were "somewhat" or "very" interested.

Patients gave doctors and other health care providers high marks for providing clear explanations on health care issues, but lower marks for dealing with patient's emotions and feelings, time spent with patients, and providing enough opportunities to ask questions. Patients said that 92% (76/83) of providers "usually" or "always" explained things in a way patients said they could understand, and 88% (73/83) "usually" or "always" made sure the patient understood what they needed to know to take care of their health. However, patients reported that 23% (19/82) of providers "never" or only "sometimes" gave enough attention to their feelings and emotions, helped them to deal with feelings of uncertainty, or spent enough time with them during the clinic visit. Twenty-three percent (19/83) of patients said that providers "never" or only "sometimes" gave them enough time to ask all of the health-related questions they had.

Patients were asked about their preferred setting to learn about their health and what types of information they were most interested in receiving. Fifty-three percent (36/69) stated they would prefer to receive this information during their visit to the clinic, either before (28%, 19/69) or after (25%, 17/69) seeing their provider. Forty-seven percent (33/69) of patients preferred reviewing clinical information at home either on their own time (17%, 12/69), before (14%, 10/69), or after (16%, 11/69) their appointment. When asked to select one or more of 6 types of health information, patients were most interested in receiving information on specific health issues (78%, 64/82), test results (68%, 56/82), medications and side effects (59%, 48/82), general health and wellness (55%, 45/82), chronic pain management (38%, 31/82), and community health resources (33%, 27/82).

Patient Interest and Attitudes Toward the Proposed Tablet-Based System

Patients expressed a high level of interest in the proposed tablet-based system, with 64% (54/84) saying they were "extremely" or "very" interested and only 5% (4/84) saying they were "not interested at all".

Compared to other methods to disseminate health information, patients believed the tablet system would be more motivating, engaging, and informative than printed materials, websites, and emails. For example, compared to printed materials, the tablet system was considered more motivating (80%, 66/83), engaging (86%, 69/80), and informative (78%, 63/81). However, the tablet system was not considered to be superior to face-to-face information exchange with providers –patients were about equally divided in choosing between the tablet or providers as more motivating (53%, 44/83 choosing tablets), more informative (51%, 40/79 choosing providers), and more engaging (54%, 43/80 choosing providers).

Patients believed the proposed tablet system would have positive effects on their health care. The majority of patients agreed that using the tablet system during clinic visits would improve their knowledge of their medical condition (60%, 49/81), would assist them in making healthy lifestyle choices (57%, 47/82), and help them to feel more comfortable talking with their provider about their medical condition (55%, 45/82). Patients were less sure of whether the tablet system would improve their relationship with their provider. Forty-four percent (36/82) agreed that the tablets improve patient-provider relationships, but 33% (27/82) neither agreed nor disagreed with this statement.

The primary concern patients expressed were regarding privacy issues. Thirty-three percent (27/81) said they were concerned about privacy using the proposed tablet system. Typical responses to an open-ended question about privacy issues were concerns of "who will have access to my information" and people "hacking into the system."

Provider Interviews

The characteristics of providers and their description of the types of patients they treat are presented in Table 2. Providers interviewed were a diverse group, consisting of 2 physicians (ie, internal medicine, general practitioner), a psychiatrist, a nurse practitioner working in a primary care clinic, and a registered nurse who directs a wellness center. Three providers were located at the same community health center where patients were recruited. Four of the 5 providers treat a diverse patient population with the internal medicine physician noting that the patients they see are mostly older, well-educated, women.

Table 2. Provider sample characteristics and the types of patients they see.

		Provider 1	Provider 2	Provider 3	Provider 4	Provider 5
Practice		Internal medicine	General practitioner	Psychiatrist	Nurse practitioner, primary care	Registered nurse, wellness center director
Clinical setting		Wellness center	Community health center	Wellness center	Community health center	Wellness center located at a community health center
Patients						
	Medical conditions	Stress-related medical conditions	Diverse, chronic conditions	Range of psychiatric conditions, many with severe depression	Diverse, chronic conditions	Chronic conditions and chronic pain
	Age, years	40-60	Adults	Adults	Adults	Adults
	Sex	Female	Female	Female	Diverse	Female
	Race/ethnicity	Diverse	Diverse	Diverse	Diverse	Diverse
	Education	Higher levels	All levels	Clinic practice, all levels; private practice higher levels	All levels	Lower levels
	Income	All levels	All levels	Clinic practice, all levels; private practice higher levels	All levels	Lower levels

A majority of providers interviewed (4 of 5) reacted enthusiastically to the idea of incorporating tablets for health information exchange into their clinical workflow. Physicians reported that they spent nearly 25-50% of their time counseling and educating patients on individual lifestyle and behavioral modifications. A majority of providers (4 of 5) are using traditional printed materials as a means to disseminate health communication materials to patients and thought the tablet system would enhance multiple aspects of the clinical encounter. Providers thought that using the tablet system would allow for more personalized content delivery with the possibility of using easy-to-understand, modular audio-visual material. Furthermore, they thought that this content would prompt patients to think about their health issues prior to their clinical encounter and allow patients more time to assimilate their health information with additional context provided by the physician as needed.

Given that data collected with the tablets could be used in the patients' longitudinal EHR, providers were also interested in using the tablets to gather patient information as a part of the clinical history or intake exam. For example, Provider 3 suggested that the tablet system could incorporate validated visual analog scales to monitor depressive symptoms, which could then be use to track the efficacy of therapy. This would contribute clinically valuable patient-generated health data to the patient's record as a measure of outcomes. Additionally, providers indicated that the flexibility of such a platform could be used to provide tailored health information content through the use of disease or medical condition specific modules.

The primary barriers with using tablets in the clinic as perceived by providers centered on 3 areas: usability by various patient populations, assurance of patient privacy, and the physical maintenance of tablets in the clinic. One provider was concerned that using a technology-based educational medium would be difficult for certain patients, particularly those who were elderly, had lower socioeconomic status and literacy, and those who were recent immigrants. Additional concerns were for users of the tablet system with movement disorders, chronic pain, and difficulties with vision/hearing, which would prevent them from effectively using the tablet interface. Providers also expressed a need to ensure patient privacy. For example, Provider 5 suggested the use of privacy screens and personal headphones for audio/visual content. Finally, physically keeping the tablets in the clinic as well as ensuring their distribution and return along with ancillary devices (headphones, chargers, privacy screens, etc) was a potential difficulty in implementation, creating additional tasks for support staff.

Discussion

Principal Findings

The goal of this study was to explore patients' and providers' attitudes regarding a proposed tablet-based personalized health care information exchange system in the waiting room and during the clinical encounter. We found that patients and providers were receptive to the idea of using tablets. A majority of patients believed the tablet system would have positive effects on their health knowledge, assist in making decisions regarding their health, and help them to feel more comfortable communicating with their provider. Patients thought the proposed system would be more engaging, motivating, and informative than other communication channels. Providers thought the tablet system would enable more personalized delivery of health education content to patients and the collection of patient-reported data for use during the clinical encounter. Barriers expressed by both patients and providers were concerns regarding the privacy and security of information collected using a tablet system.

Engaging patients in their health and medical care is understood to be key to achieving better health outcomes and higher patient satisfaction with care [35-37]. However, efforts to improve patient engagement in clinical settings are labor intensive and difficult to implement [38]. A point of care tool that delivers information, prompts patients to take action or change behaviors, and supports patient-provider communication and shared decision making may be the most effective model to improving engagement [38]. The time patients spend in the waiting room is an opportunity for them to learn about their health and make decisions regarding their medical treatment [39]. We found that patients spend about 20-25 minutes in the waiting room and that only 1 in 4 makes use of this time to learn about their health. Additionally, about half of patients preferred to receive health information just before or after their appointment, rather than outside of the clinic. This makes the waiting room an excellent opportunity to provide patients with personalized health information and implement patient-centric interventions into the clinical workflow.

Patients responded positively to the proposed tablet system and thought it would help them to improve their knowledge, assist in making decisions about their health, and feel more comfortable communicating with their providers. A similar tablet system to the one we proposed was found to have a positive effect on patients' likelihood of asking questions of their providers. Hess et al [40] found that patients randomized to the group using the tablet to receive personalized health information and feedback were more likely to ask questions about mental health issues and a larger, but not statistically significant, percentage initiated some type of discussion with their provider, compared to controls. Unfortunately, a limitation of this pilot study is the cross-sectional design in which patients were asked about their thoughts regarding the proposed tablet system, rather than interventional study design in which subjects' use of a tablet system is assessed over time. Additional research is thus needed to examine how repeated use and reinforcement of educational information and decision aids delivered by a tablet system might affect patient knowledge and patient-provider interactions.

Efforts to improve patient engagement most often employ educational materials to increase patients' knowledge and to support shared decision making between patients and providers [41]. Although these materials have been found to be efficacious in improving patient engagement and shared decision making [42], few patients receive these materials during a primary care visit. A 21-month study of 5 primary care practices in California found that only 10% of eligible patients received targeted educational materials about screening tests [41]. The primary barrier cited by physicians that limit the use of educational materials and patient-provider discussions on treatment decisions is time constraint [41]. Interestingly, Lin et al found that physicians who made greater use of educational materials as decision aids reported that it saved them time because patients could review the information before the clinical encounter. The providers we interviewed thought the tablet system would be helpful in delivering personalized information and prompt patients to think about their health and medical treatment outside of the clinical encounter. Additionally, providers were interested in the capability to collect patient-reported outcomes while in the waiting room. A tablet system could automate the data entry, scoring, and analysis of this data, which could then be used to guide the clinical encounter and populate the EHR. This capability may improve workflow efficiency and increase the amount of face-to-face time between provider and patient [16]. Tablets used in an inpatient setting have been found to reduce the time required to check the EHR and increase the time providers spend with patients [18].

Providers expressed some concerns about the feasibility of using tablets in the clinic. One concern was coordinating the distribution and return of equipment. Key to the successful integration of a tablet-based system in the clinical workflow is the adoption by the nonphysician clinic staff who will most likely be responsible for distributing, collecting, and maintaining the tablets. Previous research examining the distribution of decision support aids designed to improve patient engagement, increase knowledge, and support shared decision making-the same goals as those of the proposed tablet system-found that clinic staff were willing to take on this task and were most effective in distributing materials to patients [41]. Successful implementation will also require educating staff and providers on the benefits of a tablet system and how it benefits patients, providers, and contributes to achieving long-term institutional goals. Pilot studies that examine integrating the tablet system into the clinical workflow will be needed to understand how to mitigate barriers if the program is to be scaled successfully.

A second concern of providers was that some patients may not be able to make effective use of tablet-based systems. Research has found that patients who are older and with lower levels of income and education have more difficulty using tablets [25]. However, we found that patients were optimistic about their ability to use the proposed tablet system. Most patients were very or extremely confident they could use a tablet and greater than 90% (73/80) were willing to learn how to use one if they had to. Only 5% (4/84) of patients expressed no interest at all in using the proposed system. Other evidence suggests that concerns about older adults' use of technology are diminishing. A study investigating the use of tablets among older adults recovering from cardiac surgery found no evidence that older adults were "technophobic"—unwilling or unable to use the tablets [43]. A recent Pew report finds that older adults are rapidly adopting some types of technology, and although this group lags other segments of the population on some types of technology use, they are more likely to own a tablet than a smartphone [44]. This report also echoed the concerns raised by the providers we interviewed; that aging and health-related limitations in sensory and cognitive functioning can make using technology difficult. Continued research and development efforts are needed to ensure that new HIT systems are accessible and usable by patients despite physical and cognitive limitations.

A concern raised by both patients and providers was the issue of privacy and security. These concerns may negatively impact patient and provider trust and adoption, and poses a risk to the success of a tablet system, and more broadly, to the expanded use of HIT [45]. Privacy and security concerns are often noted as a barrier to HIT adoption [46,47]. However, patients may be less concerned about privacy than providers and are generally

willing to use technology when they view the benefits as outweighing the risks [48]. For example, in a study using focus groups to explore patients' attitudes on technology's future role in health care, researchers found that patients were less concerned about privacy and more concerned with making use of technology to improve their access to relevant medical information, communicate with their provider, and making data available to providers in an emergency [49]. Similarly, despite 33% (27/81) of patients expressing concerns about privacy, we found that 64% (54/84) were extremely or very interested in using the proposed tablet system. Ensuring high levels of trust and the success of a tablet system will require additional research to understand patients' and providers' concerns and building a platform that meets technical standards and regulations.

Limitations

Although the methods used in this study were adequate for collecting data to explore the potential for creating a tablet-based health information system for use in primary care settings, it is not without limitations. First, patients were recruited from only 1 site, a community health center affiliated with a large teaching hospital in the northeast. Although this site was selected due to the ethnic and socioeconomic diversity of the community and patients, this setting is not necessarily representative of patients, physicians, or clinics in the United States. A second limitation of this study is the small sample size which limits the coverage of all types of patients and thus, the generalizability to all patients who receive care at the clinic. Third, findings on provider attitudes are based on a very small sample of 5 providers recruited using a nonprobability, snowball sampling methodology, and should not be interpreted as representative of physicians across specialties, clinics, or regions in the United States. Finally, patients and providers responded based on an illustration and explanation of a proposed tablet system and provided initial reactions to this concept. Use of a functioning system, with its own unique features and user experience, would elicit much different responses.

Ultimately, the goal of this research is to develop and implement new HIT to improve patient engagement and self-management of patient medical conditions, while also improving the efficiency and effectiveness of health care delivery. Future research should support an "agile development" process to guide building a tablet system. In support of this goal, future research is needed to move beyond the results of this exploratory study to understand the specific features and functionality patients and providers require in a tablet system. This could take several stages. First, additional formative research is needed to collect data on these features from a more diverse sample of patients and physicians. Second, after identifying key features, these should be confirmed across potential users using questionnaires and sampling methods to collect data that are representative of the patients, providers, and the clinics and communities they serve. Third, wire-frame mock-ups of the tablet application, or early stage "alpha" versions should be developed and tested with small groups of patients and providers. Through an iterative process of development, testing, and redesign a "beta" version of the tablet application can be tested in a small feasibility study and if successful, in a larger study to evaluate effectiveness.

Conclusion

Patients and providers were highly amenable to integrating tablets into the clinical experience, and it may be useful in improving patient-provider communication, patients' health knowledge, and the collection of PROMs. Further research into operationalizing such systems and their validation with patient outcomes is necessary before integration into standard clinical practice.

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Conflicts of Interest

None declared.

Multimedia Appendix 1

Survey Instrument.

[PDF File (Adobe PDF File), 138KB - resprot_v4i4e116_app1.pdf]

Multimedia Appendix 2

Provider Interview Guide.

[PDF File (Adobe PDF File), 40KB - resprot_v4i4e116_app2.pdf]

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Abbreviations

AV: audio-visual
CHI: Connected Health Innovation
EHR: electronic health record
GDP: gross domestic product
HIT: health information technology
MGH: Massachusetts General Hospital
PROM: patient-reported outcome measure

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Original Paper

Virtual Nursing Intervention Adjunctive to Conventional Care: The Experience of Persons Living With HIV

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Abstract

Background: Persons living with HIV (PLHIV) must adhere optimally to antiretroviral therapy (ART) on a daily basis and for their lifetime to maintain an undetectable viral load, allowing them to preserve their health. Taking advantage of the opportunity that information and communication technologies provide to broaden intervention modalities and intensify clinical follow-up, a virtual nursing intervention consisting of four interactive computer sessions was developed to empower PLHIV to manage their ART and symptoms optimally. Compared with other types of information and communication technologies-assisted interventions such as text messages, HIV Treatment, Virtual Nursing Assistance and Education (VIH-TAVIE) requires a certain degree of active engagement on the part of the user to develop and strengthen the self-management skills to optimize adherence. After the intervention's impact on ART adherence was measured quantitatively, a qualitative study was undertaken to describe how users experience the intervention. Understanding how PLHIV perceive being assisted asynchronously by a virtual nurse was of particular interest.

Objective: The objective of the study was to explore and describe how PLHIV experience VIH-TAVIE, that is, receiving customized asynchronous accompaniment via a virtual nurse.

Methods: A qualitative study was conducted with 26 PLHIV (20 men, 6 women) who received all four VIH-TAVIE sessions. Participants had been diagnosed with HIV 14 years earlier on average and had been on ART for a mean period of 10 years. The sessions lasted 20-30 minutes each and were received two weeks apart. They are hosted by a virtual nurse who engages the user in a self-management skills-learning process for the purpose of treatment adherence. Semistructured interviews were conducted lasting 30-40 minutes to get participants to share their experience of the intervention through personal stories and what they thought and felt during their participation. Data were analyzed using Miles and Huberman's method, by performing these three steps: (1) data reduction (data coding, summaries); (2) data display (in tables and text form); and (3) recontextualization of results.

Results: Content analysis yielded five themes regarding how PLHIV experience VIH-TAVIE: (1) exposure to the virtual nursing intervention; (2) virtual nurse humanizes experience of the computer-delivered intervention; (3) learner's experience of the virtual nursing intervention; (4) perceived benefits following participation in the virtual nursing intervention; and (5) relevance of the virtual nursing intervention in relation to the medication management trajectory.

Conclusions: Analyzing the participants' experience revealed they found the intervention's content and format appropriate. To them, the virtual nurse humanized the experience and helped them acquire new skills for achieving optimal ART adherence. Results seem to underscore the importance of offering the intervention to persons who have more problems with drug intake or who are just beginning ART.

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KEYWORDS

medication adherence; anti-HIV agents; Internet; HIV infections; nursing research; qualitative research; web-based interventions

Introduction

Achieving Optimal Treatment Adherence With Antiretroviral Therapy

The emergence of antiretroviral therapy (ART) turned HIV infection into a chronic disease. The aim of ART is to attain and maintain an undetectable viral load, enabling persons living with HIV (PLHIV) to maintain their health. To this end, daily antiretroviral drug intake must be adhered to throughout one's lifetime. In a meta-analysis of 84 studies from 20 countries, Ortego [1] showed that only 62% of PLHIV adhered to ART optimally. This suggests a need for intervention to help PLHIV achieve optimal treatment adherence. In this regard, a recent review of interventions aimed at optimizing ART adherence highlighted five types of intervention with evidence-based efficacy: cognitive-behavioral interventions, education, treatment supporters, directly observed therapy, and active adherence reminder devices such as mobile phone text messages [2].

Text message is the most common type of intervention assisted by information and communication technologies (ICT) used to optimize treatment adherence among PLHIV, and the most tested, as reported by Pellowski and Kalichman [3] in their systematic review of ICT-based programs. Moreover, in their recent meta-analysis, Finitsis et al [4] suggested that text message intervention could help sustain ART adherence. However, some researchers, for example [5,6], have developed and tested computer- or Web-delivered educational and cognitive-behavioral interventions in this regard. These are generally complex and long-term customized health programs that patients can access repeatedly at their convenience. For example, Fisher et al [5] tested a Web-based HIV medication adherence intervention called "Life Windows" composed of several educational modules. This intervention is animated by a virtual animated character. There is no mention if this virtual guide is a health care professional. Their protocol analysis revealed a significant increase in self-reported adherence by the group that received the Web-based intervention. Hersch et al [6], for their part, evaluated a Web-based program named "Life-Steps for Managing Medications and Stress", which was comprised of nine educational and cognitive-behavioral modules. The Web-based program is fully audio narrated and encompasses video vignettes and other interactive components. Though they reported that the adherence rate among participants who received the program decreased slightly, it declined from about 85% to 66% in the control group. Robbins et al [7] tested a computer-based intervention called Masivukeni on antiretroviral adherence and key psychosocial outcomes among 55 nonadherent South African HIV patients. This intervention includes a multimedia component, where a lay counsellor delivers the intervention to help people living with HIV in resource limited settings achieve optimal adherence. The proportion of participants who achieved 80% adherence or greater, based on the clinic-based pill counts, was 67% among Masivukeni participants and 16% among standard of care (SOC) participants at post intervention (around 5 to 6 weeks post baseline). Participants in the intervention group showed significantly more positive attitudes regarding disclosure and medication social support, less social rejection, and better clinic-patient relationships than SOC patients.

HIV Treatment, Virtual Nursing Assistance and Education

We developed and tested a virtual intervention called HIV Treatment, Virtual Nursing Assistance and Education or *VIH-TAVIE* (from its French version, *Virus de l'immunodéficience humaine - Traitement assistance virtuelle infirmière et enseignement*) to empower PLHIV to manage their ART and symptoms optimally. VIH-TAVIE consists of four interactive computer sessions hosted by a "virtual" nurse who leads the user through a learning process geared toward acquiring the requisite skills for treatment adherence. What distinguishes this intervention from others is the interaction with a "virtual" nurse, a real nurse that supports and coaches PLHIV in an asynchronous way through video. This nursing intervention aims to reproduce the caregiving relationship (a consultation, a relational practice) between nurse and patient. The videos of a nurse are used to humanize the Web (computer)-based intervention, generating the illusion that someone is there to encourage the patients in their treatment-taking behavior, rather than having exclusively scripted or written content or audio media.

The Web-based interventions cited previously [5-7] have a written audio and may have video components, but there are no "real" health care professionals supporting the learning process. At times, there may be an animated character acting as a counsellor, but it is not the equivalent of having a real health care professional that embodies her professional role in a virtual context. Compared with text message interventions, VIH-TAVIE demands a certain degree of active engagement on the part of the user in order to develop and strengthen the self-regulatory skills required to deal with difficult situations as they arise. The intervention was evaluated in a hospital setting as an adjunct to conventional care. The results of this quasi-experimental study comparing the effectiveness of two types of follow-up—conventional versus conventional plus adjunctive virtual (VIH-TAVIE)—in promoting ART adherence among PLHIV revealed that both interventions improved adherence [8].

Describing the Participants' Experience

In addition to implementing quantitative approaches to measure the intervention's impact on ART adherence, we considered it was also important to describe the users experience with such an ICT-assisted intervention. There is little known about the experience of the participant in Web-based interventions designed to foster antiretroviral taking behavior. In the past, interventions such as Fischer [5], Hersch [6], and Robbins [7], did not address this in a qualitative study. In the current study, we were particularly interested in understanding how PLHIV perceived being assisted asynchronously by a virtual nurse. The

qualitative design of the study provides insights into the views of participants regarding their interaction with a virtual health professional and about the timing or dosage of the intervention. These participant perspectives may provide leads for effectively implementing the virtual nursing intervention, VIH-TAVIE, on a larger scale.

Methods

Study Design

An exploratory qualitative study based on conventional content analysis [9] was chosen to describe how PLHIV experienced the virtual nursing intervention. This method is inductive and avoids using predetermined categories to allow for a better understanding of a phenomenon.

Setting and Participants

The study took place at a university hospital that provides care and services to PLHIV. PLHIV were invited to participate if they were at least 18 years old and had been on ART for at least six months. Pregnant women, people with an uncontrolled psychiatric condition, and active intravenous drug users were excluded from the study. To be included in the qualitative section of the study, participants had to have completed all four VIH-TAVIE computer sessions and had to be able to share their experience in this regard with ease. Of the 99 PLHIV who received the virtual intervention, 73 completed all four sessions. For the purposes of this qualitative study, a convenience sample was used to explore experiences during the intervention. To achieve data saturation, 26 participants who completed all four sessions were recruited.

The study was approved by the Institutional Review Board of the Research Center of the Centre Hospitalier de l'Université de Montréal. Written consent was obtained from the participants after they were provided with a full verbal and written explanation of the study and advised that they could withdraw from the study at any time. Furthermore, the participants were assured that measures would be taken to ensure confidentiality throughout the research and protect their identity, including use of pseudonyms and modification of data collected during interviews (eg, workplace, physician's name).

Exposure to Intervention

VIH-TAVIE consists of four interactive computer sessions 20-30 minutes long. These are hosted by a virtual nurse who engages the user in a self-management skills-learning process. The sessions target different skills: self-assessment, motivational, problem-solving, emotion-regulation, and social. These enable PLHIV to integrate the therapeutic regimen in their daily routine, manage side effects, handle problem situations that might interfere with drug intake, interact with health professionals, and mobilize their social network. The sessions follow a predefined sequence in order to ensure a gradual transmission of skills [10].

At the heart of the application is a virtual nurse coach who interacts with the user asynchronously (Figure 1 shows this). Over the course of the sessions, approximately 140 video clips of the virtual nurse are presented ranging in length from 10 to 90 seconds. She delivers tailored teaching following an algorithm based, for example, on the skills performed by the user or the user's adherence level. Aside from the tailored teaching, the virtual nurse also refers to the experiences of other PLHIV who have coped successfully with situations similar to those of the user (Figure 2 shows this). During the sessions, the virtual nurse provides feedback and positive reinforcement on the user's personal style and methods and on skills acquired. Each interactive session is distinct from the other in terms of message, strategies, skills, questions, and data entry. The intervention makes different tools available to help remember new information and skills and to have specifically tailored advice at hand. To this end, the tools in question can be printed out as PDF files. They are intended as support for using the newly acquired skills and, if desired, to keep track of progress. For example, a self-observation behavior chart (Figure 3 shows this) is available for users to identify and analyze the circumstances surrounding a lapse by answering questions such as: "What is the context in which the lapse occurred?", "What was I doing?", "Who was I with?", "What was I thinking about?", and "What can I do to prevent the situation from recurring?". Some documents offer advice on how to manage side effects (Figure 4 shows this), while others are meant to facilitate drug intake. The latter tools recommend making use of positive image association and keeping a journal of side effects, and provide guidance regarding negative emotions, communication strategies, and social support. Finally, many other information documents can be accessed under the frequently asked questions (FAQ) section of the VIH-TAVIE Web interface [11].

This virtual nursing intervention is grounded in a disciplinary perspective, which is the McGill nursing model [12,13], and by extension, the strength-based approach [14]. With this approach, the person and his family are perceived as active participants in their health care and learn new ways to cope with difficult life events [12,13]. The virtual nurse supports the person in developing and reinforcing skills to manage difficulties by helping them to mobilize their strengths and their resources [15]. Bandura's self-efficacy theory [16] was used, in particular to reinforce the individual's confidence or capacity in managing their treatment/therapy, because self-efficacy was a main focus of the intervention. The virtual nurse aims to develop and reinforce self-management skills in individuals.

Figure 1. Screenshot of HIV Treatment, Virtual Nursing Assistance and Education (Virus de l'immunodéficience humaine - Traitement assistance virtuelle infirmière et enseignement) (VIH-TAVIE): interaction with the virtual nurse.

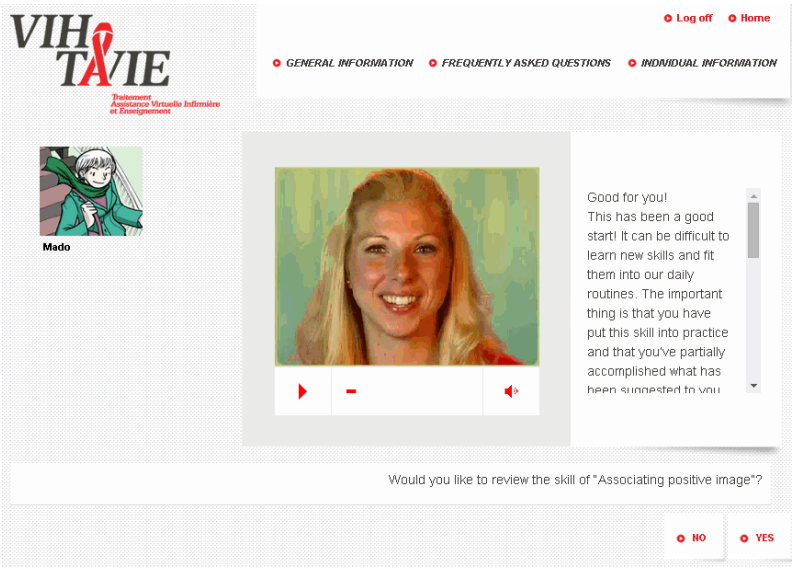
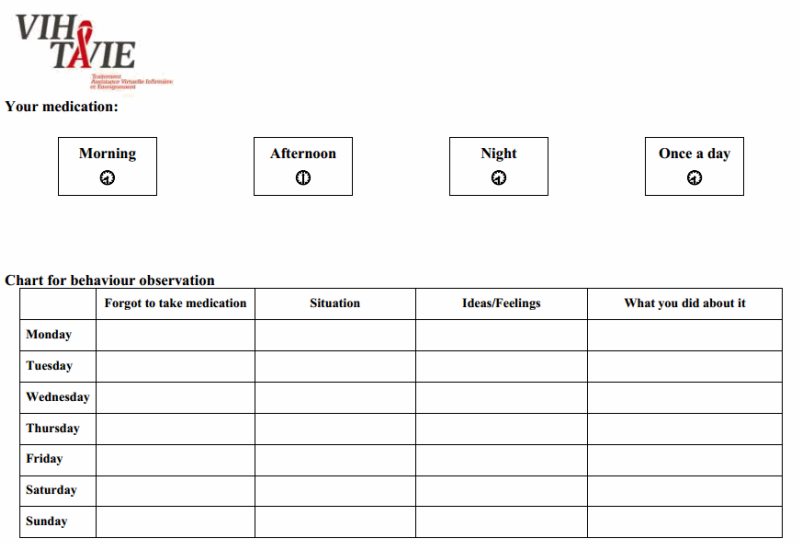


Figure 2. Experience of a person living with HIV (PLHIV) who has successfully overcome barriers. HIV Treatment, Virtual Nursing Assistance and Education (Virus de l'immunodéficience humaine - Traitement assistance virtuelle infirmière et enseignement) (VIH-TAVIE).

Figure 3. Chart for behavior observation. HIV Treatment, Virtual Nursing Assistance and Education (Virus de l'immunodéficience humaine - Traitement assistance virtuelle infirmière et enseignement) (VIH-TAVIE).



VIH TAVIE

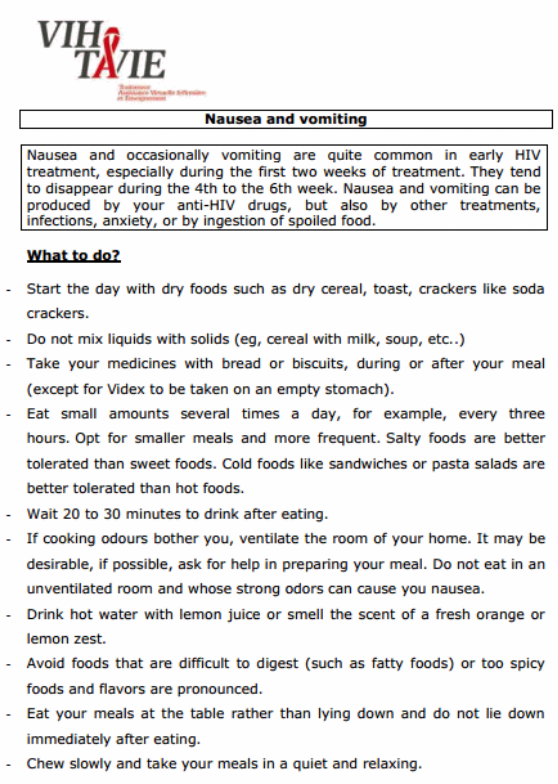
Your medication:

Morning	Afternoon	Night	Once a day
◔	◑	◔	◔

Chart for behaviour observation

	Forgot to take medication	Situation	Ideas/Feelings	What you did about it
Monday				
Tuesday				
Wednesday				
Thursday				
Friday				
Saturday				
Sunday				

Figure 4. Advice on how to manage side effects. HIV Treatment, Virtual Nursing Assistance and Education (Virus de l'immunodéficience humaine - Traitement assistance virtuelle infirmière et enseignement) (VIH-TAVIE).



VIH TAVIE

Nausea and vomiting

Nausea and occasionally vomiting are quite common in early HIV treatment, especially during the first two weeks of treatment. They tend to disappear during the 4th to the 6th week. Nausea and vomiting can be produced by your anti-HIV drugs, but also by other treatments, infections, anxiety, or by ingestion of spoiled food.

What to do?

- Start the day with dry foods such as dry cereal, toast, crackers like soda crackers.
- Do not mix liquids with solids (eg, cereal with milk, soup, etc..)
- Take your medicines with bread or biscuits, during or after your meal (except for Videx to be taken on an empty stomach).
- Eat small amounts several times a day, for example, every three hours. Opt for smaller meals and more frequent. Salty foods are better tolerated than sweet foods. Cold foods like sandwiches or pasta salads are better tolerated than hot foods.
- Wait 20 to 30 minutes to drink after eating.
- If cooking odours bother you, ventilate the room of your home. It may be desirable, if possible, ask for help in preparing your meal. Do not eat in an unventilated room and whose strong odors can cause you nausea.
- Drink hot water with lemon juice or smell the scent of a fresh orange or lemon zest.
- Avoid foods that are difficult to digest (such as fatty foods) or too spicy foods and flavors are pronounced.
- Eat your meals at the table rather than lying down and do not lie down immediately after eating.
- Chew slowly and take your meals in a quiet and relaxing.

Data Gathering

Individual semistructured interviews lasting on average from 30 to 40 minutes were conducted to allow participants to share their thoughts and feelings during the virtual nursing intervention. Few interviews lasted more than one hour. An interview guide was used to ensure all topics of interest were covered, including use/appropriation of technology, relevance of intervention, such as drug intake support, possible improvements to the intervention, and the interaction with the virtual nurse. The interview guide was developed by the research team that has an expertise on virtual intervention, with PLHIV, or qualitative research (JC, GR, PRG, AB). The questions reflected the objectives and the components of the virtual nursing intervention. The interview guide was refined during the data collection. The following are examples of questions asked: "Looking back on the VIH-TAVIE virtual intervention, what did you find least helpful?", "What did you find most helpful?", "What did you think of your contact and interactions with the virtual nurse?", "Could you describe your experience during the computer sessions?", and "Over the computer sessions, how did you feel?". Allowing the participants to freely express their experiences in their own words contributed to

obtaining a fuller description of the experiences. All the interviews were recorded and transcribed verbatim.

Data Analysis

The data were analyzed using the Miles and Huberman's method [17], whereby three activities were performed concurrently: (1) data reduction (data coding, summaries); (2) data display (in tables and text form); and (3) recontextualization of results. Each interview was coded by a principal coder. The codes were then validated by another person well acquainted with the project, and by means of consensus differences and discordances were resolved. The codes were examined for similarities and differences and grouped into categories. For instance, data associated with the virtual nurse constituted a group or category. Data capturing differences were classified under the same theme and an explanation for the differences was provided. In order to foster a more in-depth analysis, these similarities and differences were described in text form. The document was then sent to the rest of the research team for discussion. Various cycles of analysis and discussion took place, allowing for a refinement of the results and for extracting themes and subthemes relevant to the study's purpose. These results were then recontextualized and held up against the existing literature. An iterative process was followed throughout the analysis, going back and forth between the transcripts and the interpretation of the results.

The research assistant and the research coordinator (GR) undertook the following measures to ensure the credibility and authenticity of the study, and to establish confidence in the study [18]: read the interviews multiple times; alternated between data collection and analysis; reached data saturation; and used peer review and held debriefing sessions with all the members of the research team (JC, AB, PRG). They took part in some discussions regarding data collected, analysis, and interpretation. Working documents allowed the research team to discuss the emerging themes, and to interpret the findings. This iterative process was helpful to draw a portrait of the experience of interest.

Results

Sample Characteristics

The sample consisted of 26 participants who completed all four sessions. Of these, 20 were men and 6 were women. Their mean age was 49 years (SD 8; range: 32-74) and 58% (15/26) had a high school education. There were 58% (15/26) that had annual income of less than CAD $15,000. They had been diagnosed with HIV 14 years earlier on average and had been on ART for a mean period of 10 years (see Table 1).

Table 1. Sociodemographic characteristics (N=26).

Characteristics		Participants' data
Age (years), mean (SD), (min-max)		49, (8), (32-74)
Gender, n (%)		
	Male	20 (77)
	Female	6 (23)
Ethnicity, n (%)		
	Canadian	23 (88)
	Other[a]	3 (12)
Marital status, n (%)		
	Single	16 (62)
	Married or living as a couple	3 (12)
	Divorced/widowed	7 (27)
Sexual orientation [b] , n (%)		
	Heterosexual	10 (38)
	Homosexual	14 (54)
	Bisexual	1 (4)
With children, n (%)		
	No	18 (69)
	Yes	8 (31)
Education level, n (%)		
	Primary	1 (4)
	Secondary	15 (58)
	College	8 (31)
	University	2 (8)
Annual income CAD, n (%)		
	< $14,999	15 (58)
	$15,000-$24,999	7 (26)
	$25,000-$34,999	2 (8)
	> $35,000	2 (8)
Employment status, n (%)		
	Working/student	4 (15)
	Retired/unemployment insurance	2 (8)
	Welfare	13 (50)
	Other	7 (27)
Living arrangements, n (%)		
	Living alone	12 (46)
	With partner/family, friend	11 (42)
	Other (housing)	3 (12)
Years of HIV infection, mean (SD), (min-max)		14 (7.45), (0.50-26)
Years on ART, mean (SD), (min-max)		10 (5.67), (0.50-22)

[a]Other countries, South America 1, 4; others 2, 8

[b]Missing data, 1, 4

[c]Missing data, 1, 4

Themes

The analyses yielded five major themes that describe the experience of having participated in a virtual nursing intervention in support of ART adherence. These themes and their corresponding subthemes are presented below.

Exposure to the Virtual Nursing Intervention

Under this theme, the ergonomics of VIH-TAVIE and the presence of an actual nurse on site were explored as subthemes.

The Ergonomics of the Virtual Nursing Intervention

Though a small number of participants had technical difficulties associated with forgetting their log-in password, navigating the Web pages, or experienced stress when using a computer, the majority of the participants considered VIH-TAVIE to be user-friendly and enjoyable, and found it generally easy to navigate the interface,

> *I'm not familiar with computers, but it was great, it was explained very clearly[...]It went very well. It really wasn't complicated.* [Participant #5, 10 years of ART]

The audiovisual content of the VIH-TAVIE intervention, such as the images, choice of avatar, video clips, and colors, was appreciated and made participation in the virtual intervention more captivating,

> *The colors are relaxing, they're not aggressive. The colors, but also the choice of graphics was very nice[...]The window in which the virtual nurse is framed is not too big, it's not full screen, which means you don't feel invaded by her[...]Same with the graphics because they're not overdone, they're relatively simple.* [Participant #23, 13 years of ART]

Presence of an Actual Nurse on Site to Facilitate Transition to Virtual Mode

Several participants reported being reassured by the presence of an actual nurse on site before the start of the sessions, providing reassurance for the intervention with the virtual nurse. There was one person that underscored that having an actual nurse on site favored participant engagement in the intervention,

> *Had I been told simply to visit the site [VIH-TAVIE], I would have. But would I have stuck with the program to the end? I don't know.* [Participant #23, 13 years of ART]
>
> *I just felt safe having the same nurse there all the time[...]Seeing how it was the same person welcoming me at the door and, then, there she was on the screen, well, it was reassuring.* [Participant #26, 11 years of ART]

Moreover, having a person nearby during the session was helpful for those who had little or no familiarity with computers, as they could refer to the on-site nurse when questions regarding the content emerged or they required help navigating,

> *That was very encouraging. Yeah. There's always someone there with you if you have a problem or something, a button. She sorts it out.* [Participant #9, 15 years of ART]

Virtual Nurse Humanizes Experience of the Computer-Delivered Intervention

Numerous persons had the feeling of being accompanied and of interacting synchronously with a nurse, as though they were using a Webcam. Yet, the interaction modality was asynchronous via video clips of the virtual nurse. The "human" and "warm" dimension of the intervention was emphasized by nearly all the participants. The virtual nurse was pleasant to listen to and watch. She provided clear explanations, tools, and tricks essential for drug intake,

> *It feels like you're interacting. Uh, you don't get the impression it's a robot talking to you, you have the impression it's someone explaining things clearly. I might add that the visuals of the virtual nurse give you the impression she's real. She teaches you, she explains information, she gives, passes on information.* [Participant #15, 1 year of ART]
>
> *I found the project...more personal with the virtual nurse than if it had just been a computer because it felt like I was with a person. I found that interesting.* [Participant #1, 7 years of ART]
>
> *The whole thing of interacting with a virtual nurse, well, it makes me smile because...it's less cold than simply reading information and interacting with a good computer program that responds to you[...]It's less cold. So, finally, the reassuring side I was talking about, well, that's it in a way. There's someone there talking to you. It's not a computer, it's...it's human.* [Participant #13, 13 years of ART]

Learner's Experience of the Virtual Nursing Intervention

All of the participants appreciated the quality of the teaching delivered by the virtual nurse. In fact, the information presented in VIH-TAVIE was described as relevant and complete, as evidenced by the following transcript excerpt,

> *My experience with VIH-TAVIE was very worthwhile, very interesting, very rewarding[...]I must say in all honesty that it was very complete. Uh, it really covered all the bases. Uh[...]and, yeah, like it or not, in a way it forces you to question yourself all the time[...]* [Participant #16, 16 years of ART]

The majority of the PLHIV considered the questions easy to understand, but a minority found some questions lacked clarity or were repetitive. Some participants expressed feeling stressed while answering the questions and wanted to give their responses some thought to ensure their accuracy,

> *Trying to answer as best possible, as, uh, correctly as possible in terms of what corresponded to me[...]I felt that at times the answers to the questions didn't come to me fast enough* [Participant #12, 21 years of ART]

This desire to provide the correct answer is a testament to the participants' engagement in the virtual intervention.

Another positive point was the possibility of consulting the fact sheets (eg, skills summary, FAQ, advice regarding side effects) available on the VIH-TAVIE. These allowed the PLHIV to gain a better understanding of their side effects. These sheets were used as reference tools throughout the intervention and even after its completion. They allowed the PLHIV to consolidate, review, and sustain the learning achieved during the course of the sessions. The sheets were perceived as reassuring and helpful. This was the case even for participants who had been on ART for a long time. Since they were customized to their needs and accessible, the sheets empowered them to take charge of their condition.

> *I don't know what I'd do without them [fact sheets]. I'd probably be over at my doctor's all the time asking: "What do I do about feeling tired all the time? What do I do about my lack of appetite?" Instead, with all the fact sheets, I can get by on my own[...]I took all of the sheets so as to help myself. And, uh, these tools have been a great help. I mean, I was very happy to have them because, see, I'm still using them today.* [Participant #12, 21 years of ART]

Participants also appreciated the "novelty" of dealing with medication via a computer-delivered intervention. For some persons, learning to use a computer was seen as an accomplishment,

> *It's a computer-based experience education-wise. Plus, you learn a bunch of stuff...working a computer, it's important. You know, I didn't know how to use a computer, but I learned* [Participant #9, 15 years of ART]

Perceived Benefits Following Participation in the Virtual Nursing Intervention

Emotional Benefits

Completing the virtual nursing intervention allowed the participants to dampen their fears and their stress level regarding possible changes in medication. The intervention gave them confidence, reassured them, and encouraged them to continue with their ART. Acceptance of the disease and not feeling alone were also reported as other emotional benefits,

> *It cheered me up because I realized I wasn't in the same, uh...I wasn't the only one to have come down with this thing, it cheered me up. You know, I mean, listening to what she has to say about taking my medication[...]* [Participant #12, 21 years of ART]

> *It's funny but knowing that I wasn't the only one made me feel like, ok, I'm not here alone, I come here [VIH-TAVIE sessions] to improve my quality of life, like so many others who come here as well...So, how did I feel? I felt welcomed.* [Participant #21]

There was one participant that even referred to the intervention as "virtual therapy". A sense of pride emanated from these experiences of having managed to regain control of their intake behavior. Here is an eloquent quote from a participant who reported being nonadherent prior to the intervention and for whom the benefits of having participated in the intervention were irrefutable, *Truth is, this experience saved my life.*

Using New Strategies/Skills

The participants reaped benefits from the information received and the tools and tricks proposed to foster optimal antiretroviral drug intake during the intervention. For most of them, the intervention was successful in answering their questions, particularly those regarding side effects, and directed them toward resources, such as community groups. The following example is illustrative of what various participants had to say in this regard,

> *It allows us, right, to develop strategies, ok, specific to each person, because each person has their own strengths and weaknesses[...]I found that the program offered me strategies, tools, gave me a lot of tools to make sure that I take my medication as prescribed[...]Plus, I have to say that the name of the program [the acronym VIH-TAVIE is the homophonic equivalent of "live your life" in French], uh...well, I find that it's fitting in that the program allows us to live our life, uh, in a wholesome way and, yeah, fully and in good health. And to enjoy a good quality of life.* [Participant #15, 1 year of ART]

Finally, the participants reported that, thanks to the intervention, they had gained a better understanding of the reasons for their lapses, acquired means to avoid them, and developed insight on how they took their medication. Many of them indicated that, prior to the intervention, they did not always grasp why they lapsed so often,

> *But [during the intervention] I thought about my medication intake, that's for sure. About better ways of taking it. And it made me reflect also, uh, uh, on when I, of the times when I didn't take it, why I didn't take it. Because sometimes you don't always give it much thought.* [Participant #4, 11 years of ART]

One of the benefits for PLHIV, therefore, was to have gained awareness of the importance of regular medication intake, which could translate into the application of strategies to optimize intake.

The PLHIV reported having used strategies proposed by the virtual nurse to manage side effects, to adopt a positive attitude toward medication, to try out visualization techniques, and to communicate with their health professionals (eg, physician). Many of the PLHIV reported having planned out a schedule for drug intake and having incorporated it in their routine,

> *Thanks probably, precisely to the information that I got at the computer, I was able right away to put it into practice, and then set and manage a suitable schedule to make it easier, precisely, to take my medication without it becoming a burden.* [Participant #16, 16 years of ART]

Relevance of the Virtual Nursing Intervention in Relation to the Medication Management Trajectory

Relevance Depends on Quality of Prior Antiretroviral Drug Intake

The participants described the relevance of VIH-TAVIE in relation to the quality of their drug intake behavior prior to the start of the computer sessions. In this regard, the higher the reported optimal drug intake, the lesser the effects of VIH-TAVIE appeared significant to participants. For example, for one participant who claimed to be adherent prior to the intervention, and, therefore, qualified his drug intake as regular, the intake behavior was maintained over time,

> *Before VIH-TAVIE? No different. I took my medication all the time, regular. I didn't have any problems, none.* [Participant #5, 10 years of ART]

For another participant who only rarely neglected to take his medication prior to VIH-TAVIE, there was some improvement,

> *I think that, before, I used to skip my medication a little more often. Now, I almost never do. It happened once this month, but it was only the first time in, in four months, I think. It's not much.* [Participant #4, 11 years of ART]

Moreover, for the three persons who previously reported being nonadherent, participation in the virtual nursing intervention enabled them to become aware of their situation. They realized that their behavior was not optimal owing to the numerous times they neglected to take their medication. Thanks to the intervention, they identified factors that interfered with their drug intake, such as precedence given to alcohol over medication and an unstable living environment (eg, poor housing). The intervention helped these persons overcome these difficulties and adopt optimal drug intake by reducing their lapses considerably,

> *Thanks to VIH-TAVIE, I take my medication now, it's automatic[...]Look, I didn't take my medication regularly before, I skipped doses all the time. Then I did four sessions at the computer, I saw the nurses[...]Well, bottom line is it works. That's what matters.* [Participant #8, 14 years of ART]

Some participants considered that the virtual intervention helped them change other health-related behaviors,

> *Since doing those sessions...virtual, practical, written. Well, things have changed. Ok, I still like the occasional beer or a little toke. But what's that? For me it's nothing compared to before. I used to do coke every day. I'd shoot up, the whole shebang...Uh, everything available on the market I'd do. But now, those days are over. Done[...]Thanks to you, uh, to your programs and to...I read and reread, uh, my questions, the answers, everything in the kit that the nurse left me.* [Participant #10, 15 years of ART]

Timing of HIV Treatment, Virtual Nursing Assistance and Education Relative to the Medication Management Trajectory

Most of the participants recognized the relevance and added value of the intervention as drug intake support. However, the large number of participants who had been on treatment a very long time felt that VIH-TAVIE would have more of an impact if offered to persons beginning therapy or having problems with drug intake,

> *I find the program effective for people who are starting medication rather than people who have been on it a long time[...]It's effective for people who are either starting medication or have problems taking it[...]* [Participant #4, 11 years of ART]

For one participant who had been on ART for 8 years, VIH-TAVIE was timely, given that drug intake had its ups and downs,

> *Well, obviously [over the course of the VIH-TAVIE sessions I hoped] to improve my medication intake because I was lax in this regard[...]so it just so happened that the study come along at a time when, well, it's not for nothing that I did this. Because, before, I was always quite regular and then I was like, there came a time where I was like, well, you become jaded.* [Participant #17, 8 years of ART]

Discussion

Main Results

Our study describes the experiences of PLHIV who took part in the VIH-TAVIE. There were five themes that emerged regarding their experience: (1) exposure to the virtual nursing intervention; (2) virtual nurse humanizes experience of the computer-delivered intervention; (3) learner's experience of the virtual nursing intervention; (4) perceived benefits following participation in the virtual nursing intervention; and (5) relevance of the virtual nursing intervention in relation to the medication management trajectory.

Following coaching, the benefits perceived by the participants relate to skills and strategies that foster ART adherence. These include self-observation and lapse management skills, and other skills for integrating drug intake into the daily routine and managing side effects more effectively. Thus, the expected health behaviors and strategies/skills targeted by the intervention translated into gains/benefits for this service user group [11]. The coaching proposed by the virtual nurse was designed toward developing and consolidating skills requiring a sustained effort and greater engagement on the part of the user compared with interventions that primarily involve intake reminders. The development of skills during the intervention seems to contribute to improved self-efficacy as participants perceived greater self-confidence with antiretroviral drug intake going forward. As pointed out by Bandura [16], we believe that this sense of self-efficacy probably modified their cognitive processes and their emotional states. This phenomenon might explain why participants were less fearful of changes in medication, felt less isolation, and seemed to cope with the illness better.

In our study, it appears that the patients were able to engage and benefit from the intervention and perceived it as relevant. A theme that emerged was the intervention's relevance or added value with respect to quality of drug intake and intake trajectory. According to the participants, the VIH-TAVIE intervention has a greater impact if offered to persons initiating therapy and to those with intake difficulties. This perception emphasizes the importance of meeting the "felt" needs of PLHIV and to ensure the timing of intervention. This information will be taken into account when the intervention is implemented, in order to target PLHIV more effectively.

While developing the intervention, it was important to simulate the interaction between the virtual nurse and the patient. To this end, a nurse played her professional role through an audiovisual modality, hence a "virtual nurse". The idea was to develop an intervention that transcends the technology (machine) and allows for interacting with a virtual professional. The results suggest that the virtual nurse humanizes the experience of a computer-delivered intervention. In this regard, Barnard and Sandelowski [19] proposed to examine the nature of the "relationship" between two seemingly irreconcilable entities, such as technology (eg, reproductive techniques, imaging technologies) and humane nursing care. A parallel can be drawn between ICT-assisted interventions such as VIH-TAVIE and the humanization of care. According to these authors, the determinant of whether technology dehumanizes and depersonalizes is not the technology per se, but the manner in which it is employed and operationalized in specific contexts, the meaning attributed to it, and how individuals or cultural groups define what is "humane". Sandelowski [20] discussed the notions of presence (being there physically with patients) and place (space), which must be redefined for nurses in the era of technology (cyberspace). Nurses and media designers wish to create a comfortable environment to interact that is intimate and "real" [21]. It is in the nurses' interest to create a climate of shared space where the sense of being with patients is delivered through the technology.

The intervention was developed as an adjunct to clinical care. The study participants made it clear that it was important for the virtual intervention to fall within a care trajectory. With respect to the first theme, "exposure to the virtual intervention", they expressed that the on-site nurse facilitated the transition to the virtual nurse and this face-to-face contact made them feel at ease with the virtual nurse. There was one participant that found that being accompanied by an actual nurse on site fostered engagement in the intervention. In a systematic review by McDermott and While [22], the utilization of computer technology to promote self-management of chronic illness in health care settings was found to hold great promise, with the potential to change both health behaviors and clinical outcomes among chronically ill patients.

The VIH-TAVIE virtual intervention is a customized health program that provides tailored content, but also virtual coaching and educational tools to develop the skills required to enact health behaviors. The third theme, "learner's experience", brought out the PLHIV's appreciation for the quality of the content and the relevance of the information received, as in managing side effects. In a review by Linn et al [23] of the effectiveness of ICT-assisted interventions in enhancing patient adherence to prescribed long-term medication (n=13), most of the interventions reviewed were moderately or highly sophisticated tailored interventions.

Implications for Research and Practice

We propose that further research be done using a mixed-method design to capture the overall value of this type of virtual nursing intervention in different contexts. Based on the participants feedback, it would be relevant to evaluate the efficacy of VIH-TAVIE among PLHIV who have difficulties with their antiretroviral intake (ie, those who are referred by their health care professionals; and those who have a detectable viral load and high level of CD4 count) as well as those who begin the antiretroviral medication.

Since we plan to implement VIH-TAVIE for all PLHIV and to offer them an additional and complementary service as support in their treatment taking behavior, our qualitative results will be taken into account in implementing strategies. In doing so, VIH-TAVIE must be embedded into clinical practice, as part of the routine of health care professionals.

Limitations

Despite the benefits that users experience from receiving the VIH-TAVIE virtual nursing intervention gleaned from this study (ie, receiving customized accompaniment via a virtual nurse asynchronously), its single focus on participants who had completed all four computer sessions is a limiting factor.

The use of a maximum variation sample to explore various experiences during the intervention, namely, to collect data among participants who completed one, two, or three computer sessions would have greatly enhanced the study. The experience of a participant who completed only one session is likely different from one who finished the entire virtual nursing intervention. In addition, some participants completed the interview immediately after the fourth computer session of VIH-TAVIE, while other PLHIV did the interview after full participation in the study (three months after completion of the fourth computer session). It appeared easier for the participants to report on their experiences in a virtual nursing intervention immediately following the final computer session. For the purposes of this qualitative study, we were not guided by the principle of data saturation. We chose participants according to a convenience sample, which might be a limitation of our study. In return, we are confident that data saturation has been reached because no new information was obtained [24]. The motivation for using this methodology was to gain a better understanding of the experience of the persons who engaged in the intervention. In addition, this user group made up the majority of the sample (74%, 73 of 99 participants).

Conclusions

In summary, by analyzing the participants' experience, we reveal that they found the intervention's content and format appropriate. In their eyes, the virtual nurse humanized the experience and helped them acquire new skills for achieving optimal antiretroviral drug intake. Moreover, this experience seemed to modify the participants' cognitive and emotional

processes and to boost their self-confidence with respect to drug intake. Finally, the results seem to underscore the importance of offering the intervention to persons who have more problems with drug intake or who are just beginning ART. In closing, advances in ICT present an opportunity to offer adjunctive services that fit within the continuum of care. ICT constitute an indispensable means of meeting the current challenges of care accessibility and continuity.

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Conflicts of Interest

Granting of licensing options for marketing VIH-TAVIE following study completion.

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Abbreviations

ART: antiretroviral therapy
FAQ: frequently asked questions
HIV: human immunodeficiency virus
ICTs: information and communication technologies
PLHIV: persons living with HIV
SOC: standard of care
VIH-TAVIE: HIV Treatment, Virtual Nursing Assistance and Education (Virus de l'immunodéficience humaine - Traitement assistance virtuelle infirmière et enseignement)

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Original Paper

A Self-Regulation eHealth Intervention to Increase Healthy Behavior Through General Practice: Protocol and Systematic Development

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Abstract

Background: Chronic diseases are the principal cause of morbidity and mortality worldwide. An increased consumption of vegetables and fruit reduces the risk of hypertension, coronary heart disease, stroke, and cancer. An increased fruit and vegetable (FV) intake may also prevent body weight gain, and therefore indirectly affect type 2 diabetes mellitus. Insufficient physical activity (PA) has been identified as the fourth leading risk factor for global mortality. Consequently, effective interventions that promote PA and FV intake in a large number of people are required.

Objective: To describe the systematic development of an eHealth intervention, MyPlan 1.0, for increasing FV intake and PA.

Methods: The intervention was developed following the six steps of the intervention mapping (IM) protocol. Decisions during steps were based upon available literature, focus group interviews, and pilot studies.

Results: Based on needs assessment (Step 1), it was decided to focus on fruit and vegetable intake and physical activity levels of adults. Based on self-regulation and the health action process approach model, motivational (eg, risk awareness) and volitional (eg, action planning) determinants were selected and crossed with performance objectives into a matrix with change objectives (Step 2). Behavioral change strategies (eg, goal setting, problem solving, and implementation intentions) were selected (Step 3). Tablet computers were chosen for delivery of the eHealth program in general practice (Step 4). To facilitate implementation of the intervention in general practice, GPs were involved in focus group interviews (Step 5). Finally, the planning of the evaluation of the intervention (Step 6) is briefly described.

Conclusions: Using the IM protocol ensures that a theory- and evidence-based intervention protocol is developed. If the intervention is found to be effective, a dynamic eHealth program for the promotion of healthy lifestyles could be available for use in general practice.

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KEYWORDS

intervention mapping protocol; physical activity; fruit intake; vegetable intake; eHealth; self-regulation; general practice

Introduction

Chronic diseases, such as cardiovascular disease, type 2 diabetes, and cancer, are the principal cause of morbidity and mortality worldwide, representing 68% of all deaths [1]. An increased consumption of vegetables and fruit reduces the risk of hypertension, coronary heart disease, stroke, and cancer. Furthermore, an increased fruit and vegetable (FV) intake may also prevent body weight gain, and therefore indirectly affect type 2 diabetes mellitus [2]. Insufficient physical activity (PA) has been identified as the fourth leading risk factor for global mortality [3]; it accounts for 6% of all deaths per year, and accounts for 21-25% of breast and colon cancers, 27% of diabetes, and 30% of ischemic heart disease worldwide [4,5]. Consequently, effective interventions that promote PA and FV intake in a large number of people are required. Reviews have shown that eHealth interventions are effective in changing PA and FV intake [6,7]. In eHealth interventions, information and communication technologies are used to improve or enable health and health care [6]. eHealth interventions allow a personalized approach at a relatively low cost by making use of interactive, computerized technologies [6,8], and have several advantages: reduced personal demands, consistency over time, increased interactivity and flexibility, automated data collection, and more honest self-report. Most of these interventions are delivered through the Internet only [6,9,10]. Although a large number of people can be reached through the Internet, the percentage of individuals who start with an Internet-delivered intervention is low, and sustained use is even lower [11]. Reach and use of eHealth interventions can be enhanced by the provision of additional support [11]. General practitioners (GPs) may be influential in supporting patients by providing extra information when implementing the intervention [12-14]. The reach and sustained use of eHealth interventions may also be increased by using computer-tailored feedback and facilitating goal setting and self-monitoring, as well as by incorporating email and short message service (SMS) text message reminders as behavior change methods into eHealth interventions [15-17]. Although eHealth interventions have been shown to be effective, effect sizes of eHealth interventions that target PA and dietary behavior are small [16,18]. Computer-tailored feedback merely targets variables that primarily address the adoption of an intention to change (eg, attitude or social norm), hence leaving many individuals in an intention-behavior gap. It is also important to address this gap by addressing postintentional factors (eg, action planning and problem solving). A self-regulation perspective may be well suited to integrate both pre- and postintentional processes, and to develop interventions that guide individuals during all phases of behavior change [19,20]. Self-regulation techniques can empower adults and allow them to make more autonomous decisions about their own health behavior [19,21,22]. Self-regulation is a goal guidance process which occurs in a motivational and volitional phase [19]. During the motivational phase (ie, goal selection, goal setting, and representation), participants become aware of risks, form intention-to-change behavior, and set goals to change their behavior. In the volitional phase (ie, active goal pursuit, goal attainment, and maintenance or goal disengagement), participants make action plans, engage in goal pursuit, and maintain or adapt their goals [19].

In this study, a dynamic eHealth intervention, MyPlan 1.0, was developed that targets self-regulation processes to increase PA and FV intake. To enhance reach and use, the intervention will be implemented in general practice. To ensure that MyPlan 1.0 is theory and evidence based, as well as feasible for implementation in general practice, the intervention mapping (IM) protocol was used as the planning model for the intervention [23]. IM facilitates effective decision making by formalizing the development process of the intervention in six steps [23]. Via the IM protocol, researchers are guided in selecting target behaviors (Step 1), specifying intervention goals (Step 2), choosing intervention strategies (Step 3), and developing intervention tools and programs (Step 4). IM also involves the planning of the implementation (Step 5) and the evaluation (Step 6). This paper describes the theoretical considerations and decisions made during each step of the IM protocol. This resulted in a detailed intervention description, which provides insight into the design and different components of the intervention, and will help planners to identify techniques and replicate the different intervention components [24].

Methods

The IM protocol consists of the following six steps: (1) needs assessment, (2) development of matrices of change, (3) selection of theory-based methods and practical applications, (4) description of the program production, (5) development of a program adoption and implementation plan, and (6) completion of an evaluation plan [23].

In Step 1, a planning group was established to ensure that the intervention targets important factors to increase effectiveness and sustained used of the eHealth intervention. Based on the needs assessment, we also selected the target behaviors in Step 1.

In Step 2, we adopted self-regulation theories to determine the intervention content and to formulate performance objectives. Different statements were formulated about how participants may achieve the intervention goals. These statements are specific actions that have to be taken by participants and are called performance objectives [23]. Next, relevant and changeable determinants of the target behavior were selected in the second step. Finally, change objectives were formulated by stating what needs to be changed regarding a determinant in order to accomplish a performance objective.

In Step 3, theory-based methods that can modify the selected determinants to achieve the performance objectives were determined. Matching methods were selected based upon the results of systematic reviews that summarized the effectiveness of behavior change methods for healthy eating and physical activity interventions. We also took into account the summary list published by Bartholomew et al [23] and the taxonomy of behavior change techniques published by Abraham and Michie [24].

In Step 4, an intervention plan was developed, based on the selected methods and practical applications. Previous programs

that were effective were used as examples [25-28]. Furthermore, a pretest of the intervention was conducted in Step 4 to identify possible elements of improvement and to evaluate the feasibility and user-friendliness of the intervention program.

In Step 5, the implementation of the intervention was planned. Support by general practitioners has been shown to improve the use and the effect of computer-tailored programs [11]. Therefore, GPs were involved in the implementation of MyPlan 1.0 in general practice. To this end, during Step 5, we conducted focus group interviews with GPs regarding the implementation.

The aim of this paper was to describe the intervention development. It is therefore not a research protocol of the trial, which is reported at ClinicalTrials.gov (trial registration number: NCT02211040). Therefore, in Step 6, we only specify the evaluation design and briefly describe the evaluation plan. We briefly describe the decisions made during each step of the IM protocol in the results section.

Results

Step 1: Needs Assessment

The planning group consisted of six researchers from different health disciplines—physical activity, nutrition, psychology, and primary health care—and leading GPs from the Belgian association of GPs, who are potential end users of the program. The core theories, methods, practical applications, implementation options, and evaluation strategies were discussed among this planning group.

Based on needs assessment, physical activity and fruit and vegetable intake were selected as target behaviors. Insufficient physical activity and unhealthy diet are two important risk factors of chronic diseases (eg, diabetes and ischemic heart diseases) and cancers (eg, breast and colon cancer) [1,29]. Adults are recommended to have 30 minutes of moderate-intensity aerobic PA 5 days per week, or to have 20 minutes of vigorous-intensity aerobic PA 3 days per week [30]. However, these recommendations are not reached by a large part of the population [5], nor by a majority of Belgian adults (62%) [31]. Therefore, it was decided to target physical activity levels in various subdomains (eg, activities at work, activities during leisure time, and active transports and sports).

Adults are recommended to consume at least five portions or 400 grams of fruit and vegetables a day, and data from the World Health Survey showed that 78% of the adult population consumed less than five portions of fruit and vegetables daily [32,33]. Western adults (ie, in Belgium, Luxembourg, France, Ireland, the Netherlands, and Great Britain) consume on average 129 grams of fruit and vegetable per day [2]. Furthermore, an evaluation of the gap between food-based dietary guidelines and the usual food consumption in Belgium indicated that fruit and vegetable consumption was significantly lower than recommended in a large part of the Belgian population. Of Belgian adults, 53% and 62% do not eat fruits and vegetables on a daily basis, respectively [34]. Therefore, we also decided to focus on fruit and vegetable intake as a dietary component of the intervention. After the needs assessment, the intervention goals were as follows: (1) to increase fruit and vegetable intake, and (2) to increase physical activity levels in Belgian adults (older than 18 years).

Step 2: Performance Objectives, Determinants, and Change Objectives

The performance objectives for the target PA are shown in Table 1, and those for fruit and vegetable intake are shown in Multimedia Appendix 1. For example, the first performance objective for PA is “Adults recognize the importance of increasing PA levels.”

Table 1. Performance objectives for physical activity.

Phases	Performance objectives
Motivational phase	Goal selection, setting, and representation
Performance objective 1	Adults recognize the importance of increasing physical activity levels
Performance objective 2	Adults decide to change their physical activity levels and set physical activity goals
Volitional phase	Active goal pursuit
Performance objective 3	Adults choose their own strategies to change their physical activity levels
Performance objective 4	Adults start pursuing their physical activity goals
Performance objective 5	Adults monitor and evaluate their physical activity levels
Performance objective 6	Adults maintain or adapt their physical activity goals to a higher level
Performance objective 7	Adults adapt their goals and strategies when they are unable to reach their initial goals

The health action process approach model of Schwarzer [20] was used to identify and categorize determinants within a self-regulation framework. This model has been successfully applied to predict fruit and vegetable intake [35,36] and physical activity [20,37-40]. The model categorizes determinants into two phases: a motivational phase and a volitional phase [20]. In the former phase, risk awareness, outcome expectancies, and preaction self-efficacy are determinants that influence intentions. After an intention is formed and goals are set, participants try to achieve their goals. In the volitional phase, action planning, coping planning, maintenance self-efficacy, and social support are determinants that influence actual changes in fruit and vegetable intake and physical activity levels. Maintenance and recovery self-efficacy are important determinants for participants to choose to maintain or adapt their goals, based on an evaluation of their behavior change [20].

All performance objectives, related change objectives, and determinants for the target behavior PA are shown in Multimedia Appendix 1. For example, to accomplish performance objective 2—"Adults decide to change their PA level and set PA goals in one or more subdomains"—change objective 2.5—"Adults identify for which PA goals they have the highest level of confidence"—describes what needs to be changed regarding the determinant *preaction self-efficacy*.

Step 3: Selection of Theory-Based Methods and Practical Applications

In Tables 2 and 3, an overview of the methods and practical applications used in the intervention is given for the motivational and volitional phases, respectively. For example, the selected theoretical method *stating implementation intentions* corresponded with the determinants *action planning*, and *coping planning*.

To translate the methods into practical applications, we used study protocols of effective interventions [25-28,41-45]. We also used methods incorporated in an original program from our research group developed by Vandelanotte et al [27] and Spittaels et al [28]. This original program gave only feedback on motivational determinants (eg, intentions, attitudes, and knowledge). To effectively translate techniques that also target volitional determinants into practical applications, we used the programs of van Genugten et al [41], Walthouwer et al [25], and Springvloet et al [26]. For example, a practical application that was formulated by the method of *implementation intentions* to target *coping planning* was to let adults formulate a coping plan by formulating if/then plans (ie, implementation intentions). After the "if" is determined, selected difficult situations or barriers are stated. After the "then" is determined, selected solutions to overcome these difficult situations and barriers are stated (eg, *If* it is Monday evening and I am not in the mood for sports, *then* I call my friend to go to the aerobic lessons together).

Table 2. Methods and practical applications used in the intervention for the motivational phase.

Methods	Determinants	Practical applications
General information	Risk awareness	General information is provided in the form of short texts and slogans. In these texts and slogans, physical activity guidelines and health benefits of sufficient physical activity levels are highlighted.
	Outcome expectancies	Adults can read information about physical activity and select the information that they are interested in on a website. They can, for example, select to read information about positive outcomes due to sufficient physical activity levels or information about the benefits of increasing physical activity levels.
Monitoring, tailored feedback, and personal risk information	Risk awareness	After filling in a questionnaire about physical activity level, personal feedback is provided in which adults' levels of physical activity are provided, as well as how these compare to the recommended level.
Tailored feedback and modelling	Preaction self-efficacy	The tailored feedback includes stories about peers who succeeded in increasing physical activity levels, also in difficult situations. For example, "Eric (40 years old) decided to be more physically active in his free time, by walking in the local park for 30 minutes, three times per week. When it was raining, Eric decided to go swimming instead of walking."
Prompting identification of barriers and problem solving, and tailored feedback	Preaction self-efficacy	A predefined list of possible difficulties (barriers and risk situations) to increase physical activity level is provided and adults can select these difficulties that are applicable to them. Based on their answers, tailored information and tips for solutions to overcome the indicated barriers and risk situations are provided; adults can select those solutions to apply which they are confident about.

Table 3. Methods and practical applications used in the intervention for the volitional phase.

Methods	Determinants	Practical applications
Selecting hindering factors/barriers and solutions Implementation intentions	Action planning Coping planning	Adults can first select hindering factors and barriers out of a predefined list. When applicable hindering factors and barriers are not available in the list, participants also have the possibility to write down another factor or barrier in an open-ended format. Next, participants can select solutions out of a predefined list or write down another solution. Afterward, participants are stimulated to make action plans and coping plans by formulating if-then plans (ie, "implementation intentions"). After the "if," a situation or the previously selected difficult situations or barriers are stated and after the "then" the selected action or solutions to overcome the difficult situations and barriers are stated (eg, If it is Monday evening and I am not in the mood for sports, then I call my friend to go to the aerobic lessons together). Adults can formulate this implementation intention plan in an open-ended question format on the website.
Goal setting	Action planning	A list with personal and relevant goals is formed based on previous answers; adults can select the goals to change that they are confident about.
Stating SMART[a]goals	Action planning	Adults are guided by questions to make a *specific*, *measurable*, *attainable*, *relevant*, and *time-bound* (SMART) action plan. For example, adults can formulate answers to questions on what they want to do (eg, increase physical activity by biking 20 minutes to work), how often (eg, three times per week), when (eg, Monday, Wednesday, and Friday), and when they want to start (eg, starting on Monday, July 7). After answering all the questions, the personal action plan and the if/then plan are automatically generated and sent by email to the participant.
Public commitment	Social support	Adults can choose to send their action plan to others (eg, family and friends) to ask them to support them and invite them to also make an action plan.
Prompt self-monitoring of behavior and prompt review of behavioral goals	Action planning Maintenance self-efficacy	Adults are asked to keep a record of their physical activity levels or fruit and vegetable intakes by one of the given suggestions (ie, personal paper agenda, mobile phone, Excel sheet, or online agenda). After the active goal pursuit was started, adults are also invited by email to report their behavior on the website. Periodic email reminders are sent to invite adults to fill out a questionnaire about the target behavior and their goals on the website. The results are compared with their previous behavior and goals, and iterative feedback is provided on the progress of behavior change.
Set tasks on a gradient of difficulty	Maintenance self-efficacy	When adults have attained their goals, they are invited to change the goal by reformulating a more attainable or more difficult goal or by setting additional goals.
Planning coping responses	Coping planning Maintenance self-efficacy	Adults are asked whether they experienced barriers while pursuing their goals. If so, they are invited to identify solutions to cope with the identified situations or barriers. Adults can again select solutions from a list that is generated based on the selected difficulties.
Prompt review of behavioral goals and personal feedback	Recovery self-efficacy	When people do not achieve their goals, people get personal feedback that informs them that relapse is normal. They are also advised to try again, to choose other strategies, or to adapt their goals to more attainable goals.

[a]SMART: specific, measurable, attainable, relevant, and time-bound.

Step 4: Producing the Program and Materials

MyPlan 1.0 was programmed in the freely available software LimeSurvey 2.0 [46]. In what follows, the intervention program is discussed; an overview of the intervention program is given in Figure 1. MyPlan 1.0 consisted of three modules—fruit, vegetables, and PA—that are available on a website and on a tablet computer. Participants can log in, choose a behavior of interest, and run through the first session of the chosen health behaviors.

In the first session, people fill out a questionnaire and receive tailored feedback about how their behavior compares to the health norms. Next, adults can select and read more information about the behavior (eg, in relation to diseases and health) and can make an action plan. To make an action plan, adults first have to indicate whether they expect difficulties in changing their health behaviors. If so, adults can select or formulate barriers and reflect upon possible solutions to overcome the barriers. Afterwards, adults can make an if/then plan and an action plan by reading tips and filling in questions about how, when, and where they will act on their behavior. Based on the answers, an action plan is generated by the computer's algorithm (see Figure 2).

It is proposed that participants monitor their behavior when they start pursuing their goals, and are invited to send their action plan to friends or family. When session 1 is completed, the action plan is emailed to the participant. A week after adults make their action plan, they receive an email with a link to the

website where they can evaluate whether their formulated goal was accomplished. The current behavior is compared with the previous behavior and health goals, and iterative tailored feedback is provided. Based on this feedback, participants can decide to further pursue their goal, or to adapt their goal to a more difficult or more attainable goal. Participants also have the opportunity to reflect on encountered difficulties and to search again for solutions. The last session has a similar structure as session 2, and is available 1 month after completing session 1. At the end of session 3, patients are also referred to the module *Own Choice* on the website. This is an extra module, which participants can use at any time to adapt or to create an action plan for a behavior of their choice (eg, water intake). In this module, the same framework (ie, what, when, where, how many times, with who, and if/then) is used to enable adults to make a new action plan.

A further task in Step 4 is to test the feasibility, acceptability, and user-friendliness of the intervention [23,47]. Therefore, a specific study was conducted to address these issues, and its results are reported in a separate publication [48]. Briefly, 194 adults who used the MyPlan 1.0 intervention filled out an online questionnaire containing items about quality, user-friendliness, and applicability of the content and information architecture (ie, organization and delivery of the content) of the intervention. The results indicate that the program was generally well accepted, including for participants with a low educational level and for older participants. Nevertheless, to make the program more comprehensible for the different groups, the questions, answer options, and advice were made shorter and clearer [48]. To test the acceptability and user-friendliness of the tablet as a delivery mode, we conducted a thinking-aloud test with 40 adults. Most participants indicated that it was easy to use the intervention program on a tablet. Examples of comments that were reported were as follows: “text is too small to read on a tablet,” “moving from one page to another is too slow,” “and a pen to tick the answers would be useful.” Based on the comments, the intervention program was further adapted for appropriate use on the tablet.

Figure 1. Overview of the intervention program.

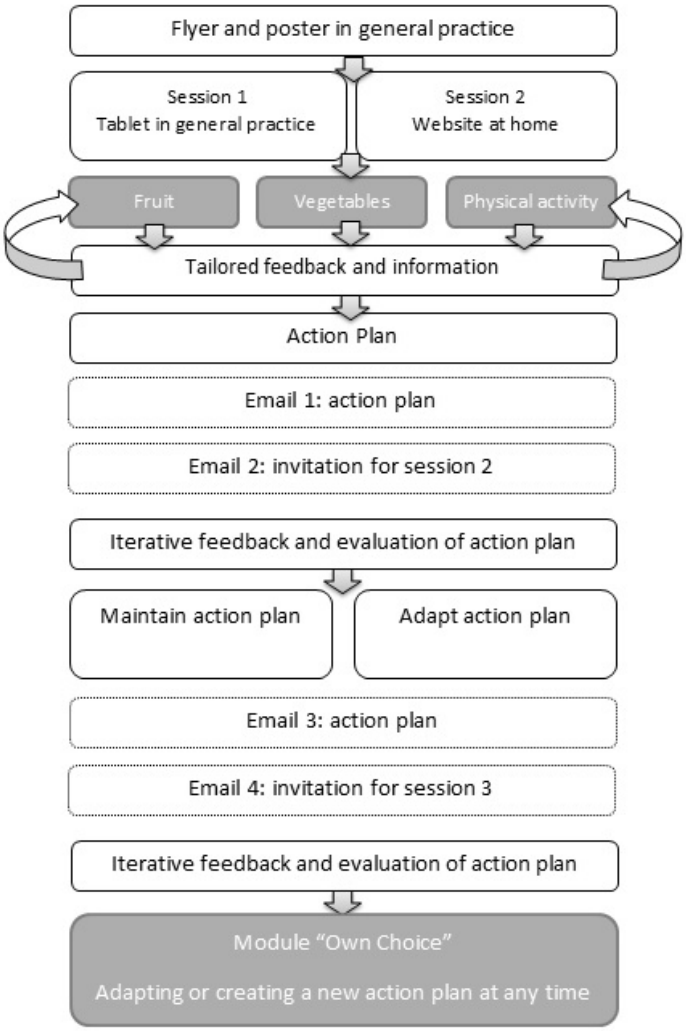
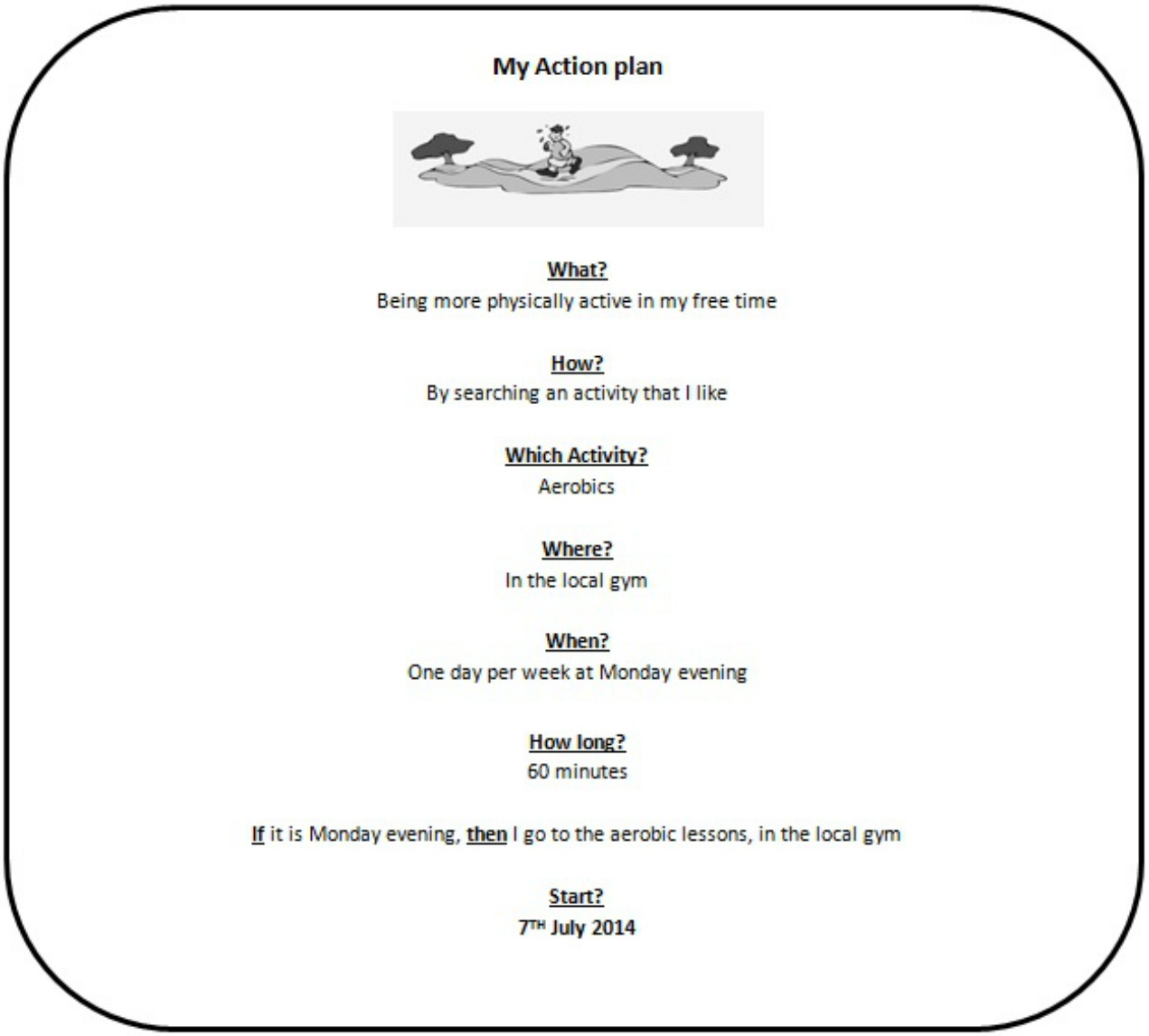


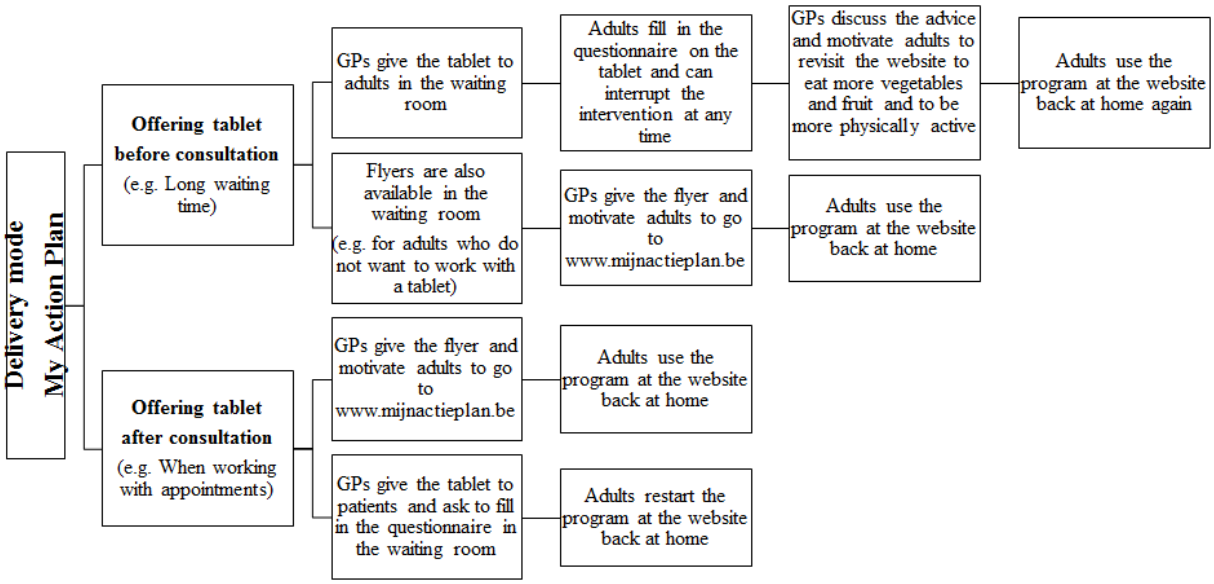
Figure 2. My Action plan: Example of an action plan for physical activity.



Step 5: Development of a Program Adoption and Implementation Plan

GPs who participated in the focus group interviews were positive about the use of a computer-tailored program that provides personal advice. GPs also appreciated that they did not need the expertise and time to compose personal advice for every patient, and may restrict their role to simply motivating and advising patients to use the intervention. However, doubts were raised on how to implement MyPlan 1.0 in general practice. By using tablets, MyPlan 1.0 patients can directly experience the use of the program and discuss their advice with their GP [49]. However, GPs indicated that in some situations it is not possible to use a tablet. For example, when there is not enough time, or when patients cannot work with a tablet. Therefore, it was decided to use a combination of flyers and tablets. Patients receive a flyer with a personal code in general practice, and can decide whether they start the program in general practice on a tablet or back at home at the website. On the flyer, it is also mentioned that participants can choose whether they want to discuss their personal advice and action plan with their GP in a following consultation. To briefly discuss the personal advice and action plans of patients, every participating GP received an information letter and attended a personal information session. In this session, GPs were instructed to emphasize the importance of personal and attainable health goals, rather than prescribing health recommendations and general information. GPs' opinions also differed about when to offer the intervention in general practice, indicating that different ways to use the program in general practice and solutions with different choices on how and when to use the program must be offered to GPs. Based on these results, it was decided to provide several modes of delivery that may be applicable in different workflow systems in general practice. Therefore, a decision tree (see Figure 3) with different choices on how to deliver the intervention in general practice was developed. In this way, GPs can autonomously decide which method is suitable for their own working system, for different patients, and for different circumstances. Other results of the focus group interviews are reported in more detail elsewhere [50].

Figure 3. Decision tree for GPs: General practitioners can use the decision tree to decide on how to implement the intervention in general practice.

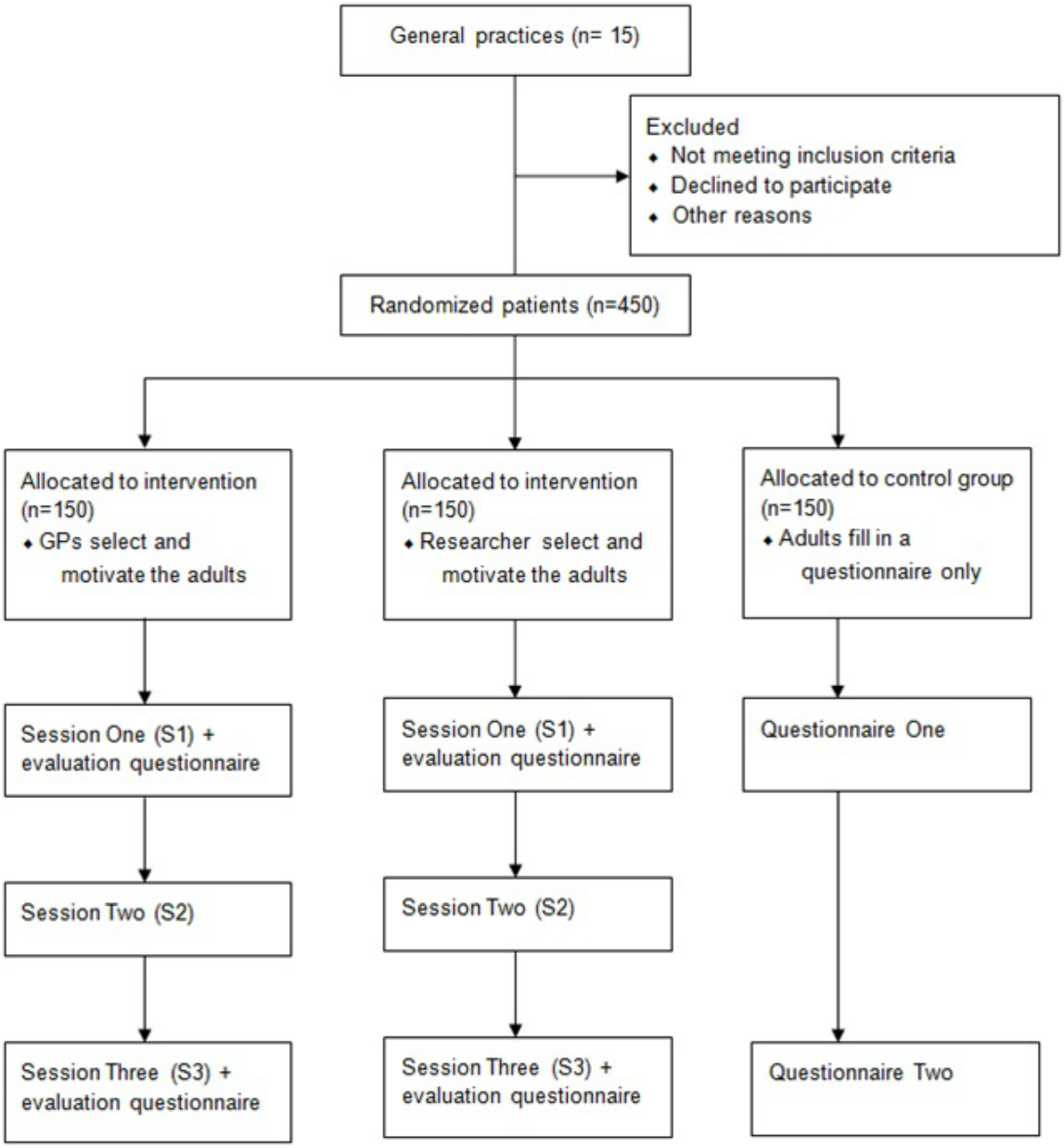


Step 6: Evaluation of the Program

A clustered quasi-experimental trial with three conditions will be used to evaluate the intervention (see Figure 4). We use a quasi-experimental design in which participants can be part of three different groups. First, a researcher recruits patients and randomly allocates them into an intervention (Group 1) or control group (Group 2). Next, a GP recruits patients into the other intervention group (Group 3). In both intervention groups, participants will not be randomly allocated to one of the behavior groups of the intervention (ie, PA, fruit, or vegetables), but participants can choose themselves for which behavior they want to complete the intervention. Group 1 is an intervention group in which researchers select and motivate adults to use the intervention by offering a flyer and/or tablet. In Group 3, the selection and motivation of adults will be randomly conducted by GPs in the waiting room by offering a flyer and/or tablet. Participants of both groups will receive a flyer with a personal code and can choose to start the intervention program in general practice on a tablet or at home on the website. Group 2 will be a waiting-list control group in which adults are randomly selected by a researcher. Participants in this control group only have to fill in a questionnaire and have no access to the computer-tailored feedback, action planning part, or to the evaluation in the follow-up modules. After completing the questionnaire at baseline and at the 1-month follow-up, the control group will also get access to the intervention modules.

In total, 30 adults will be selected in each of 15 general practices (n=450). First, a researcher will select 10 patients that will be allocated to the intervention group and 10 patients that will be allocated to the control group. Next, GPs will be asked to recruit another 10 patients to complete the intervention program. In this way, it can be evaluated whether GPs' involvement leads to more sustained use of the eHealth intervention, and higher levels of PA and FV intake. In both intervention groups, adults will be invited to complete session 1 either on a tablet in general practice or on their computer at home. Adults who do not use the tablet have to fill out a short questionnaire and leave their email address to be sent a reminder email to complete session 1 at home. After 1 week and 1 month of completing session 1, adults will receive an email to respectively start sessions 2 and 3. In the control group, adults will have to fill out a questionnaire at baseline in general practice or at home and at 1-month postintervention. To prompt adults to complete all questionnaires and sessions, reminder mails and SMS text messages will be sent. Inclusion criteria for participating in the study in both intervention and control groups are as follows: at least 18 years old, understand Dutch language, have an email address, and have access to the Internet. The outcome measures—increase in PA level, increase in FV intake, and self-regulation skills from baseline to postintervention—will be compared for the control and intervention conditions by conducting repeated measures multivariate analyses of variance (MANOVAs). Participant characteristics (ie, socioeconomic status [SES], age, sex, health status, and reaching health norms) will be compared at baseline. Characteristics that differ for the intervention and control groups will be added as covariates in further analyses. Furthermore, multilevel analyses will be conducted to take into account the clustering of participants into general practices.

Figure 4. Design of the clustered quasi-experimental trial: A clustered quasi-experimental trial with three conditions will be used to evaluate the intervention. Group 1 is an intervention group recruited by a researcher, Group 2 is a control group recruited by a researcher, and Group 3 is an intervention group recruited by a GP.



Discussion

Using IM increases the likelihood of developing an effective eHealth intervention and the transparency of intervention components, which makes replication possible for future researchers [24]. In the first step of the IM protocol we identified that PA levels and fruit and vegetable intake of adults were lower than recommended. In Step 2, the most important individual determinants for these low levels of PA and FV intake were determined and the objectives of the program were formulated. In Step 3, behavior change techniques were selected that are thought to affect these determinants and, hence, to achieve the stated objectives. In Step 4, the eHealth intervention MyPlan 1.0 was developed. Implementation strategies were selected in Step 5 and an implementation plan was made in Step 6.

Various eHealth interventions are based on motivational theories like the theory of planned behavior [51-53]. They most often target motivational determinants that are important during the early stages of behavior change, such as attitude and knowledge. However, interventions based upon theories of intentions are often more effective in changing intentions than in changing behavior [9,16], hence revealing the so-called intention-behavior gap. Our new eHealth program was partly based on a previous eHealth program developed by Vandelanotte et al [27] and Spittaels et al [28]. This original program was also based on the theory of planned behavior [54] and the transtheoretical model [55] and only gave feedback on motivational determinants (eg,

intentions, attitudes, self-efficacy, and knowledge). The strategy of tailored feedback was further integrated to target the motivational determinants, but we also searched for new strategies such as goal setting, self-monitoring, and prompt review of goal progress to target volitional determinants.

Self-regulation has recently been considered as a preferred method to overcome the intention-behavior gap and thus to promote health behavior [19,40]. Therefore, the integration of the self-regulation skills in MyPlan 1.0 that target both motivational and volitional determinants was a particular strength of our study. Previous research showed more goal ownership in participants who set their own health goals. Participants who pursue their own health goals are also less likely to drop out of behavior change programs compared to participants who get prescribed health goals [56]. Another strength is that participants have the opportunity to choose between different target behaviors and can decide themselves what they would like to change. Choice is further incorporated into the program by letting participants select for themselves what information they want to read, which goals they want to set, and which strategies they want to use. It is expected that these features will increase goal ownership, which is known to lead to more internal motivation and empowerment, and more effective behavior change [19,21,22,25,57].

A further strength of our study is the comprehensive involvement of GPs in Step 5—implementation in general practice—of the IM protocol. To ensure the feasibility of the implementation in general practice, we involved GPs from the start of the development of the intervention. During focus group interviews, important barriers for the implementation of the intervention in general practice were reported. For example, the time burden for GPs when participating in preventive actions was of major importance. Therefore, an intervention in which the personal advice was provided by a computer program was well appreciated. MyPlan 1.0 can prompt GPs to motivate their patients to adopt a healthy lifestyle, but GPs are not expected to provide extensive preventive counseling. However, some GPs will make more of an effort than others to motivate patients. Therefore, in the evaluation study all participating GPs will be asked to motivate patients to use the intervention program to set personal and attainable health goals, rather than to prescribe health recommendations and general information. Also of importance is the creation of different choices about how and when GPs may implement the intervention. Therefore, a decision tree and a list of practical solutions to implement the intervention via tablets and flyers in general practice was generated.

In Step 6—evaluation—it will be investigated whether the direct involvement of GPs in the program matters. More specifically, we will evaluate whether GPs' involvement leads to more sustained use of the intervention and higher levels of PA and FV intake. Also, multilevel analyses will be conducted to control for the clustering of participants into different general practices. Furthermore, it will also be important to evaluate the quality of participants' action plans because previous research has shown that action plans of participants can be of poor quality [58].

Following the IM protocol is a complex and time-consuming enterprise [44,59]. Therefore, we suggest that future researchers search for existing study protocols that describe the development of interventions and integrate similar theories and methods that can be used as the basis for their intervention programs. In our study, existing protocols were used to translate self-regulation methods into practical applications in a computer-tailored program [25,26,41]. However, as target behaviors and contexts differ, it is important to further elaborate the different steps of the IM protocol for new interventions. Our program, for example, differs from the existing protocols in several ways. First, we used a program that made it possible to deliver tailored follow-up feedback in which changes in health behavior were mentioned and compared with health goals. This makes it possible to provide detailed tailored feedback on the behavior change process. Second, the delivery mode of our program is variable, as the program can be delivered via different channels, such as via the Internet and via tablets. The delivery via tablets made it possible to deliver MyPlan 1.0 in natural settings (ie, general practice). This leads us to the third important aspect on which our program deviates from other programs [25,26,41], namely, the possibility to integrate extra personal feedback by general practitioners. In conclusion, if MyPlan 1.0 is found to be effective, a new eHealth program for the promotion of PA and FV intake that can be applied by GPs will be available. Future research can focus on designing modules for other behaviors and on evaluating other methods and effective channels for implementation.

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Authors' Contributions

JP developed the intervention and drafted the manuscript. IDB, GC, and MV supervised the development of the study, helped to draft the manuscript, and revised the manuscript for important intellectual content. AO participated in the development of the content of the intervention and also revised the manuscript for important intellectual content.

Conflicts of Interest

None declared.

Multimedia Appendix 1

Performance objectives, their related change objectives, and their determinants for the target behaviors physical activity and fruit and vegetable intake.

[PDF File (Adobe PDF File), 76KB - resprot_v4i4e141_app1.pdf]

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Abbreviations

CAPHRI: School for Public Health and Primary Care
FV: fruit and vegetable
FWO: Research Foundation-Flanders
GP: general practitioner
IM: intervention mapping
MANOVA: multivariate analysis of variance
PA: physical activity
SES: socioeconomic status

SMART: specific, measurable, attainable, relevant, and time-bound
SMS: short message service

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Original Paper

An Online Tailored Self-Management Program for Patients With Rheumatoid Arthritis: A Developmental Study

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Abstract

Background: Every day rheumatoid arthritis (RA) patients make many decisions about managing their disease. An online, computer-tailored, self-management program can support this decision making, but development of such a program requires the active participation of patients.

Objective: To develop an online, computer-tailored, self-management program integrated with the nursing care, as nurses have an important role in supporting self-management behavior.

Methods: The intervention mapping framework was used to develop the program. Development was a multistep process: (1) needs assessment; (2) developing program and change objectives in a matrix; (3) selecting theory-based intervention methods and practical application strategies; (4) producing program components; (5) planning and adoption, implementation, and sustainability; and (6) planning for evaluation.

Results: After conducting the needs assessment (step 1), nine health-related problems were identified: (1) balancing rest and activity, (2) setting boundaries, (3) asking for help and support, (4) use of medicines, (5) communicating with health professionals, (6) use of assistive devices, (7) performing physical exercises, (8) coping with worries, and (9) coping with RA. After defining performance and change objectives (step 2), we identified a number of methods which could be used to achieve them (step 3), such as provision of general information about health-related behavior, self-monitoring of behavior, persuasive communication, modeling, and self-persuasion and tailoring. We described and operationalized these methods in texts, videos, exercises, and a medication intake schedule. The resulting program (step 4) consisted of an introduction module and nine modules dealing with health-related problems. The content of these modules is tailored to the user's self-efficacy, and patients can use the online program as often as they want, working through a module or modules at their own speed. After implementation (step 5), the program will be evaluated in a two-center pilot trial involving 200 RA patients. Log-in data and qualitative interviews will used for a process evaluation.

Conclusions: The intervention mapping framework was used to guide development of an online computer-tailored self-management program via a process which could serve as a model for the development of other interventions. A pilot randomized controlled trial (RCT) will provide insight into the important outcome measures in preparation for a larger RCT. The process evaluation will provide insight into how RA patients use the program and the attrition rate.

Trial Registration: Netherlands Trial Register (NTR): NTR4871; http://www.trialregister.nl/trialreg/admin/rctview.asp?TC=4871 [accessed 13-NOV-15] http://www.webcitation.org/6d1ZyIoEy

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KEYWORDS

intervention mapping; eHealth; self-management; rheumatoid arthritis; tailoring; nursing care, early RCT

Introduction

Rheumatoid arthritis (RA) is a chronic autoimmune disease which predominantly affects the joints. Many RA patients face physical problems such as pain, stiffness, and fatigue which cause difficulties in everyday life [1]. RA has also been linked to depression, helplessness, and anxiety and has a considerable impact on quality of life [2-5]. As life expectancy increases and the number of people living with a chronic condition increases, there has been an increase in the number of RA patients [6].

Although healthcare professionals can give patients advice and support during visits and appointments, patients have to make day-to-day decisions about management of their disease by themselves. Self-management programs can help RA patients to take an active role in the everyday management of their disease [7-9]. Self-management has been defined as the tasks undertaken by patients to manage the symptoms, treatments, lifestyle changes, and physical and psychological consequences associated with their illness [10]. Although self-management support programs are available, most programs are provided in clinical settings or in small groups [11], and not all RA patients are willing or able to participate. With a growing number of people having Internet access and the increasing use of the Internet among RA patients [12,13], an online self-management support program can be a sustainable way to support self-management behavior. Compared to face-to-face programs, online programs provide an easily accessible opportunity to reach a large group of RA patients. Also, online programs have the possibility to tailor information and can provide more anonymity than face-to-face programs. Other advantages include 24-hour availability and avoiding waiting lists [14,15].

In the Netherlands, 2 studies, one in adolescent RA patients [16] and the other focusing on work-related problems [17], have shown that the use of self-management programs is feasible for specific groups of RA patients. At this moment, there is no generic online self-management program for adult RA patients in the Netherlands. As nurses have an important role in supporting self-management behavior, such a program should preferably be integrated in the nursing care provided as part of the multidisciplinary RA care.

An online self-management program is a complex intervention. First, it should include a variety of components, such as information provision, management of symptoms, social support, and communication strategies [18]. Second, because the target population can be diverse, self-management programs should be extensive and tailored to patient needs. Within the population of RA patients there is variance in the need for self-management support, depending for example on age, level of education, gender, or work status. Third, programs should enhance patient understanding of the behavioral change required for self-management. To develop such a program requires an understanding of the factors which influence self-management behaviors.

To ensure that our development process took account of these 3 overarching requirements, we used the intervention mapping framework. We chose to develop a tailored intervention because adapting communications and behavioral change strategies to patient needs [19] means that a higher proportion of the patients receive information that is personally relevant, which increases their motivation to change their behavior [20].

This article describes the development of an online computer-tailored program and the design of an evaluation procedure using the intervention mapping framework which could serve as a guide for the development and testing of other interventions.

Methods

The intervention mapping framework is designed to ensure that development work is focused on the most important determinants of behavior. Intervention mapping has been used successfully to develop health programs related to, for instance, medication adherence [21], promoting physical activity [22], healthy lifestyles [23], and asthma management [24]. The intervention mapping framework provides a way of systematically integrating theoretical research, empirical findings, and data collected from the population [25]. Intervention mapping provides a 6-step framework for developing health education programs: (1) identifying problem behaviors and determinants through needs assessment; (2) developing a matrix of performance objectives and change objectives; (3) selecting theory-based intervention methods and practical application strategies; (4) producing program components; (5) planning and adoption, implementation, and sustainability; and (6) planning for evaluation [25]. Active patient participation in the development process was secured by recruiting, during the first step, a multidisciplinary panel consisting of health professionals, researchers, and patients who were involved in every step of the development process.

Step 1: Needs Assessment

First we recruited a multidisciplinary panel of 5 RA patients, 2 rheumatologists, one rheumatology nurse, a psychologist, a physiotherapist, an occupational therapist, and 3 researchers (RMZ, HRW, and BvG). The rheumatology nurse and rheumatologist played a crucial role in the development and implementation of the program.

Our needs assessment comprised two components: (1) a literature search for information on health problems, problems

affecting health-related behaviors, and determinants of problems and (2) input from 2 meetings of the multidisciplinary panel. During the first meeting, we held a brainstorming session to identify the main health problems affecting RA patients. To select the most important health problems for RA patients, we coded health problems found in literature and discussed this among the multidisciplinary panel. Selection was further based on recognizability and importance of the health problems. In the second meeting, we identified problems affecting health-related behaviors and their determinants based on the literature and discussed the following questions among RA patients and health professionals: (1) why do patients have problems and (2) why do patients have problems with this behavior? In the third meeting, we asked the multidisciplinary panel whether the listed problems in health-related behavior were easily changeable or not. After these meetings, the researchers listed and coded the health problems, the problems affecting health-related behaviors, and their determinants manually.

Step 2: Developing a Matrix of Performance Objectives and Change Objectives

In the second step, we organized the performance objectives and change objectives as a matrix to indicate which behaviors needed to change to achieve the overall goal of the program, which was to enhance patients' ability to self-manage their disease and thus improve their quality of life. The performance objectives formalized the behavioral changes RA patients needed to make to achieve the behavioral goals of the program. The change objectives were performance objectives linked to changeable determinants of behavior. Thus, change objectives state what needs to change in determinants to achieve the performance objectives. Researcher RMZ constructed a matrix of the relationships between performance and change objectives which was subsequently validated by the multidisciplinary panel.

Step 3: Selecting Theory-Based Intervention Methods and Practical Applications

After defining the matrix we selected theory-driven methods on the basis of behavioral change theories. In this study, 2 independent researchers linked methods from the classification of the behavior change techniques to the problems affecting health-related behaviors and their determinants in order to select methods which could be used to achieve our overall goal. The behavior change technique classification defines strategies used in supportive programs [26]. Using a summary produced by the 2 independent researchers, the multidisciplinary panel decided whether the methods were suitable for the RA patient population. We assessed the conditions under which the methods are shown to be effective to translate methods into practical applications such as texts and videos.

Step 4: Producing Program Components

Program development was based on the change objectives and the selected theory-driven methods and consisted of composing program materials and pretesting these materials. Our research group worked with an information and communications technology partner to produce the program materials. The research group developed the content, including textual material, and our information and communications technology partner incorporated this material into an online program.

Our pretest of the online program comprised testing of the program materials by the multidisciplinary panel and testing of the program by 3 RA patients not involved in its development using the "think aloud" method [27].

Step 5: Planning for Adoption, Implementation, and Sustainability

Intervention mapping steps 1 through 4 formed the basis of the implementation. Meetings of the multidisciplinary panel were held to identify and categorize barriers and facilitators to implementation of the online program. The rheumatologist and specialist rheumatology nurse played a crucial role in the implementation process.

Step 6: Planning for Evaluation

In the final intervention mapping step we planned to evaluate the feasibility of the study design and the online self-management program by conducting an exploratory randomized controlled trial (RCT) and a process evaluation [28]. To do this we identified outcomes and process measures that were relevant to the program objectives. We also intend to conduct qualitative interviews with nurses, users, and nonusers of the program. Finally, we plan to monitor which topics related to the program components are discussed during nursing consultations and whether they are raised by the nurse or the patient.

Results

Step 1: Needs Assessment

Health Problems and the Underlying Behavioral Problems

We selected the 8 most important health problems in daily life for RA patients: pain, fatigue, stiffness, daily functioning, sexuality, work, social activities, and coping with RA.

We identified 9 general problems affecting health-related behavior from our literature review and through discussions among the multidisciplinary panel: (1) balancing rest and activity, (2) setting boundaries, (3) asking for help and support, (4) use of medicines, (5) communicating with health professionals, (6) use of assistive devices, (7) performing physical exercises, (8) coping with worries, and (9) coping with RA.

Determinants of Problem Behaviors

Our literature search determined that the following factors were relevant to problems affecting health-related behavior: knowledge, awareness, risk perception, social influence, attitude, self-efficacy, and habits. Patients confirmed the relevance of these determinants.

Step 2: Developing a Matrix of Performance Objectives and Change Objectives

The results of the needs assessment were used to draw up a matrix of performance and change objectives. One of the performance objectives was "the patient is able to set her or his

boundaries." This performance objective was relevant to the following health problems in daily life: pain, fatigue, social activities, and work.

Next we formulated change objectives relevant to the determinants knowledge, attitude, self-efficacy, and risk perception; for example, the patient knows the consequences of not setting his or her boundaries (knowledge), and the patient is conscious of the positive consequences of setting boundaries (attitude).

Step 3: Selecting Theory-Based Intervention Methods and Practical Applications

We used our matrix of change objectives to select a theory on which to base our intervention. The matrix placed the most emphasis on self-efficacy, attitude, and subjective norms. The theory of planned behavior posits that these constructs are the most important determinants of behavior, so we based our interventions on this theory. We also emphasize knowledge and awareness in our matrix, as these are preconditions for self-efficacy, attitude, and subjective norms. We then made a list of techniques which could be used to improve self-efficacy, attitude, subjective norms, and their preconditions.

For this, we derived the following methods per determinant from the coding manual for behavioral change techniques [26]. *Determinant knowledge*: provide general information about health behavior, increase memory and/or understanding of transferred information. *Determinant awareness*: risk-communication, self-monitoring of behavior, self-report of behavior. *Determinant social influence*: provide information about peer behavior. *Determinant attitude*: persuasive communications, belief selection, reinforcement on behavioral progress, providing contingent rewards. *Determinant self-efficacy*: modeling, practice, plan coping responses. *Determinant intention of behavior*: develop medication intake schedule. *Determinant action control*: use of social support, use of cues, self-persuasion. We operationalized these methods as follows: we used texts to increase knowledge, awareness, attitude, social influence, and action control; we used videos and exercises with feedback options to increase self-efficacy; and we encouraged patients to keep a diary within the online program to increase their awareness of their own health status and use an intake schedule to increase intention of behavior. We also tailored the program to the user's self-reported level of self-efficacy, because self-efficacy has been found to predict changes in various health-related behaviors [29].

Step 4: Producing Program Components

We used the change objectives and the practical applications as the basis for the online program, "Reuma zelf te lijf," which has 10 modules consisting of 2-5 sessions each. Table 1 gives an overview of the content of the modules. The first module is the introduction module and offers a short textual introduction to the other modules as well as providing information about how the program works. After this, users can respond to a series of statements; the responses are used to tailor recommendations about which module or modules users are likely to find most helpful for improving their self-management. Examples of statements include: "I want to learn to balance my daily schedule better," "I want to learn how to ask for support and help," and "I want to learn how to say no to others, for example, when I'm too tired to do something." Once users choose a module, they can work through it at their own pace, whenever they want. Every module starts with a text providing information about the topic of the module, what the patient can expect to learn from the module, and how the module is structured. Most modules allow users to respond to 2 questions to tailor the module to their self-efficacy. The responses to these questions are used to advise patients which session to move to next (session 2 for patients with a low level of self-efficacy; session 3 for patients with a high level of self-efficacy). Session 2 focuses on the following four determinants, knowledge, risk perception, awareness, and attitude, and uses informative and persuasive texts, videos of peers, and exercises to improve patients' insight into their disease and behavior and to change their attitudes. Session 3 focuses on self-efficacy and gives users the opportunity to do exercises in familiar surroundings—for example, doing an exercise to learn how to say no to others at home with a friend. Session 4 tells users how to put the skills into practice in daily life. After each exercise, users are given the opportunity to evaluate performance by responding to a set of questions. This evaluation exercise is used to help patients identify the barriers and facilitators that are relevant to their behavior. In all exercises, it is recommended that users seeks support from their partner, family, or friends. See Multimedia Appendix 1 for an example of a module.

Table 1. Overview of the modules in the online program.

Modules	Number of sessions	Topics
0. Welcome	1	Short introduction to all modules and a questionnaire to assess the level of self-efficacy
1. Balancing activity and rest	4	Planning of activities
		Keeping a balance in daily life in the long term
2. Setting boundaries	5	Dare to set boundaries (say “no”)
		Setting boundaries (communicate saying “no”)
3. Asking for help and social support	4	Establishing and maintaining social contacts
		Asking for help or support
4. Use of medicines	4	How to collect information about medication
		Taking prescribed medication
5. Communication with health professionals	4	How to prepare for an appointment with a health professional
		Asking questions and/or expressing concerns during an appointment with a health professional
6. Use of assistive devices	4	Information on how to apply for assistive devices
		Deciding whether an assistive device can help you and if so, what assistive device
7. Performing physical exercises	4	Examples of physical exercises
		How to fit physical exercises into your daily life
8. Coping with worries	3	Insight into your worries
		Controlling your worries
9. Coping with RA	2	Information and tips on how to cope with RA

During the pretest the collaborative multidisciplinary panel found that the information and the exercises provided in the modules were understandable/readable and applicable. The layout and structure of the modules were described as attractive and clear. The 3 patients who tested the program using the “think aloud” method found it difficult to navigate through the program. In response to this, we adjusted the program to make navigation easier.

Step 5: Planning for Adoption, Implementation, and Sustainability

We have planned a trial which will be conducted in 2 Dutch hospitals. The managers of the 2 rheumatology departments met regularly with the researchers to discuss trial procedures. The multidisciplinary panel identified barriers and facilitators relevant for the implementation of the online program. This information was used to design an implementation plan for the 2 hospitals which focuses on dissemination of the online program and the user's experience of interacting with the online program. We asked the specialist nurses to bring the online program to the attention of their patients during appointments. For this, a researcher explained the modules and exercises in the program to specialist RA nurses to facilitate integration of the online program with nursing care. To ensure that users’ first experiences of the program were positive, we sent potential users a written instruction manual for the program. To encourage repeated use of the program, users will be sent reminders via email.

Step 6: Planning for Evaluation

To evaluate the feasibility, we plan to do an exploratory RCT as advised by the Medical Research Council's framework for the development and evaluation of complex interventions [30]. The aims of our feasibility study will be to evaluate the potential effectiveness of the online program for patients with RA and determine effect sizes for the various outcomes, identify outcome measures most likely to capture potential patient benefits and evaluate long-term participation and attrition rates for the online, computer-tailored self-management program [31]. Because the exploratory RCT is not expected to be powered to identify differences between groups, there is no sample size calculation. Considering the complexity of the intervention and the potentially large heterogeneity of the RA population, 200 eligible RA patients will be recruited by 2 hospitals in the eastern part of the Netherlands [NTR4871]. Inclusion criteria will be diagnosis of RA, aged 18 years or older, ability to speak and read Dutch, and access to a computer with an Internet connection. Patients receiving psychiatric or psychological treatment will be excluded. RA patients will be randomized to the intervention or control group. The control group will receive care as usual; the intervention group will have access to the online program in addition to receiving care as usual.

To explore which outcome measures are most likely to capture patient potential benefits, we chose the following potential outcomes: (1) the Patient Activation Measurement (PAM-13), which assesses the knowledge, skills, and confidence for

self-management [32]; (2) the health-related quality of life survey (RAND-36), which assesses general health status in 8 dimensions (physical functioning, social functioning, role limitations [physical problems], role limitations [emotional problems], mental health, vitality, and pain [33]); (3) the Rheumatoid Arthritis Self-Efficacy scale (RASE), which measures the level of task specific self-efficacy for self-management [34]; (4) the Perceived Efficacy in Patient-Physician Interactions (PEPPI-5) [35]; (5) the short version of the self-management ability scale (SMAS-S), which measures taking initiative, investing in resources for long-term benefits, maintaining variety in resources, ensuring resource multifunctionality, self-efficacy, and maintaining a positive frame of mind [36]; (6) A scale to assess the focus on fatigue (MPCI-F) [37]; and (7) the Numeric Rating Scales (NRS), which measure pain and fatigue during the previous 2 weeks including at the moment of measurement. All instruments will be administered at baseline (T0) and after 6 (T1) and 12 months (T2). Data on the following patient characteristics will be gathered: age, gender, living situation, educational level, employment status, Disease Activity Score (DAS-28 score), physical ability using the Modified Health Assessment Questionnaire (MHAQ), time since diagnosis, current treatment, comorbidity, usage of other support programs including online programs, date of last visit to a rheumatologist, and date of last visit to a specialist nurse.

In the process evaluation we will use the framework of Saunders et al [38] to evaluate feasibility of the online program. The key components of the process evaluation are fidelity, dose received, dose delivered, reach, recruitment, and context. Data for the process evaluation will be collected from log-ins to the online program, a user questionnaire, and qualitative user interviews. The process analysis will make use of log-in data (exposure and continued use of the program), data on use of modules, and data on performance of the exercises. The user questionnaire will ask about the comprehensibility, usefulness and length of the texts and exercises, and the layout and log-in procedure. During the qualitative interviews, the frequent users and those who have stopped using the program will be asked about their reasons for using or not using the online program, which will give us insight into potential limitations and yield ideas to improve the program. We will also interview nurses to elicit their views about how introduction of the online program might affect their professional role.

Finally, to get insight into whether the online program changes the roles of patients and nurses in management of RA we have made a checklist to be completed after nursing appointments with patients in both the control group and the intervention group that covers what topics were discussed during the nursing consultation and whether it was the nurse or the patient who raised a particular topic.

Discussion

Strengths and Limitations

This article describes the systematically developed generic online, computer-tailored self-management supportive program for adult RA patients.

Using intervention mapping to structure the development process is a strength of this program. In the needs assessment, we successfully defined health problems, problems affecting health-related behaviors, and determinants of problems with health-related behavior which were relevant to RA patients. Extending the needs assessment to encompass determinants of behavior gave us a good understanding of the causes of problems with health-related behaviors. The online program uses tailored behavioral change strategies too, which should improve the likelihood of RA patients' ability to manage their disease. Another strength of intervention mapping is the use which is made of input from patients and health professionals. Integrating the experiences, knowledge, and visions of these diverse groups with scientific insights enabled us to develop a well-grounded intervention, tailored to the preferences and support needs of RA patients.

A second expected strength of the program is that the program is computer-tailored to the user's level of self-efficacy. This ensures that RA patients receive material which is suited to their personal needs, and this may increase motivation to persist with exercises and strategies recommended therein. The online format has the further advantage that patients can use the program as often as they want or need. They can choose which modules to work through and can do so at their own speed, whenever they want.

A third expected strength of the program is the extent to which the online program can be integrated with regular nursing care. All the topics covered in the program fall within the scope of a specialist RA nurse's expertise and can be discussed during appointments with a nurse. Specialist RA nurses can encourage RA patients to use the program and hopefully benefit by continuing to practice self-management.

The composition of the multidisciplinary panel might be considered a limitation of the study. All 5 RA patients had long disease duration and had found ways to cope with their illness and may not have been able to recall the problems they had in the early phase of the disease. However, in each meeting we asked the patients to try to remember how things had been when they were first diagnosed.

Another limitation might be the choice of the channel for communication relatively early in the development process. Instead of in step 4, we decided in step 1 to use an online program as communication channel. This influenced our choices at certain points in the development process, for example the choice for behavioral change strategies. This is in conflict with the concept of intervention mapping as an iterative process. However, choosing to use eHealth early on gave us the opportunity to learn about its pros and cons and how to deal with them during implementation.

Conclusions

This article describes how to develop a self-management program in a structured way and could serve as a guide for the development of similar interventions. The study yielded an online, computer-tailored self-management program suitable for all RA patients. In the planned exploratory RCT, we will assess important outcomes and estimate the relevant effect sizes;

this should be useful preparation for a larger RCT. The process evaluation will give us more insight into how RA patients use the program, which will inform future development of the program. We hope that this online self-management program will become one of the treatment options available to RA patients as part of an integrated disease management plan.

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Conflicts of Interest

None declared.

Multimedia Appendix 1

Example of a module.

[PDF File (Adobe PDF File), 90KB - resprot_v4i4e140_app1.pdf]

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Abbreviations

RA: rheumatoid arthritis
RCT: randomized controlled trial

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Original Paper

Development and Pilot Evaluation of a Tablet-Based Application to Improve Quality of Care in Child Mental Health Treatment

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Abstract

Background: Children need access to high quality mental health care. Effective treatments now exist for a wide range of mental health conditions. However, these interventions are delivered with variable effectiveness in traditional mental health service settings. Innovative solutions are needed to improve treatment delivery quality and effectiveness.

Objective: The aim of this study was to develop a scalable, sustainable technology-based approach to improve the quality of care in child mental health treatment.

Methods: A tablet-based resource was developed with input from mental health training experts, mental health providers, and patients. A series of qualitative data collection phases (ie, expert interviews, patient and provider focus groups, usability testing) guided the initial concept and design of the resource, and then its refinement. The result was an iPad-based "e-workbook" designed to improve child engagement and provider fidelity in implementation of a best-practice treatment. We are currently conducting a small scale randomized controlled trial to evaluate the feasibility of e-workbook facilitated child mental health treatment with 10 providers and 20 families recruited from 4 local community-based mental health clinics.

Results: Usability and focus group testing yielded a number of strong, favorable reactions from providers and families. Recommendations for refining the e-workbook also were provided, and these guided several improvements to the resource prior to initiating the feasibility trial, which is currently underway.

Conclusions: This study aimed to develop and preliminarily evaluate a tablet-based application to improve provider fidelity and child engagement in child mental health treatment. If successful, this approach may serve as a key step toward making best-practice treatment more accessible to children and families. As various technologies continue to increase in popularity worldwide and within the health care field more specifically, it is essential to rigorously test the usability, feasibility, acceptability, and effectiveness of novel health technology solutions. It is also essential to ensure that patients and providers drive decision making that supports the development of these resources to ensure that they can be seamlessly integrated into practice.

Trial Registration: Clinicaltrials.gov NCT01915160; https://clinicaltrials.gov/ct2/show/NCT01915160 (Archived by WebCite at http://www.webcitation.org/6cPIiQDpu)

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KEYWORDS

technology; mobile health; child mental health treatment; feasibility test; fidelity; patient engagement; traumatic stress

Introduction

Background

One in four US children experiences a mental health disorder with severe impairment or distress during their childhood [1-3]. Ensuring that these children have access to the highest quality mental health care is a top public health priority. Efficacious child and adolescent treatments exist for a wide range of mental health disorders [4]. However, these treatments are delivered with variable fidelity in mental health service settings, even among well-trained providers [5-10]. Provider fidelity generally refers to the degree to which a clinician adheres to a treatment protocol and delivers the treatment competently [11]. Drifting or deviating from empirically supported treatment protocols can diminish an intervention's potency and effectiveness, leading to a major quality shortfall [10,12-14]. Statewide and national dissemination and implementation initiatives are underway to narrow these gaps [15-17]. However, the problem of weak and inconsistent provider fidelity persists and must be addressed to improve the quality of care.

Technology-Based Resources and Access to High Quality Care

Recent technological advances offer an opportunity to support the effective delivery of best-practice interventions. This can be achieved with portable mobile apps that assist providers as they implement treatment activities that are challenging to deliver with high fidelity and child engagement. Technology-based decision support tools have been developed in the broader health care field, and initial data suggest that this approach improves clinical decision making and adherence to best practices and treatment protocols [18-19]. Novel technology-based therapy tools can offer a standardized guiding framework for providers to follow as they progress through a treatment and incorporate several design features to promote provider fidelity to a treatment model, such as (1) inclusion of a diverse yet finite set of in-session and homework activities that are all consistent with the goals of the treatment (ie, rather than off-topic or off-task activities that may encourage drift); (2) presentation of key intervention-related concepts in a consistent manner across providers and clients; and (3) assessment and tracking of progress through a treatment model for each client. Moreover, studies in child education suggest that the integration of interactive games, touch screen learning, video demonstrations, and other engaging features enhance child engagement in learning activities [20-21]. Together, these data increase confidence that technology-based approaches may have value toward improving child engagement and provider fidelity in child mental health treatment. Increased child engagement is particularly important during mental health treatment sessions, because engagement has been shown to reduce risk for dropout, which is another pervasive problem in mental health treatment that limits its impact [22].

Efforts to develop technology-based solutions for mental health care must account for limited resources available in community mental health settings, including the cost and providers' time [23-25]. Contemporary mobile devices such as tablets and mobile phones are low cost and increasingly ubiquitous [26]. Integration of these devices into practice is therefore likely to be feasible from a cost perspective. With regard to time and effort, it is important that novel solutions are user-friendly and able to be integrated readily into practice with minimal provider training and preparation. This is most likely to be achieved when providers and patients are the key drivers of the development process, working closely with the research team at each phase of design, development, and evaluation. A patient- and provider-centered approach is critical for successful implementation and dissemination.

Research is needed that directs the process of developing novel health care solutions and that measures their potential to improve the quality of mental health care. Research reviews suggest that technology-based tools, broadly considered, effectively enhance mental health care; however, the majority of this research focuses on self-help tools for adults [27-29] or other resources used by patients outside the context of formal treatment sessions [30-32]. Our protocol differs from prior approaches by examining the benefits of mobile device apps used *in session* with an emphasis on interventions designed for children and their caregivers.

Selection of a Treatment Model With Which to Test a Novel Technology-Based Solution

Selecting an appropriate treatment model is an important step in the process of evaluating a new health technology solution. Ideally, the treatment model should have a strong evidence base and high potential for cross-application with other treatment approaches to enhance generalizability of the data. We selected Trauma-Focused Cognitive Behavioral Therapy (TF-CBT) [33-34]; because it is a well-established treatment for children in the mental health field [33-38], has ample evidence supporting its effectiveness [33-38], is widely used, and has been disseminated internationally. Moreover, TF-CBT uniquely addresses multiple symptom domains commonly encountered in mental health treatment settings, including posttraumatic stress, depressed mood, and disruptive behavior. Development of a tablet-based application, or "e-workbook," for TF-CBT therefore would appear to have high potential to enhance the relevance of our data to a range of established treatments for youth. TF-CBT also requires caregiver involvement, which will allow us to explore the use of in-session resources with adults as well as children. In our pilot work, we conducted structured 30-min interviews with 21 certified national TF-CBT trainers, which revealed significant provider interest in and likely acceptance of tablet-based aids for delivering TF-CBT. These interviews also provided strong direction around key challenges that tablet-based resources can overcome [39].

Purpose and Aims

The purpose of this project was to develop and pilot a tablet-based e-workbook that is designed to increase quality of care in child mental health treatment via improvements in child engagement and provider fidelity. The potential impact of an e-workbook approach, if successful, is extraordinarily high in light of its scalability and sustainability. In the current protocol, we describe a strategy to develop an e-workbook to augment delivery of TF-CBT, a well-established treatment for children and their caregivers. If this approach is found to have utility in practice, it can be applied to a wide range of treatment approaches. The general outline of the protocol addresses three aims: (1) to develop an e-workbook to support delivery of treatment with high fidelity; (2) to conduct usability tests of the e-workbook with families and providers; and (3) to conduct a feasibility trial comparing TF-CBT vs iPad-facilitated TF-CBT.

Methods

Study Design

The aims of this investigation were accomplished in three phases. Phase I included the initial development of the TF-CBT e-workbook. Phase II included focus groups and individual interviews with 21 providers and 24 children (aged 8-16 years) to inform the refinement of the e-workbook (ie, alpha testing). It also included internal beta testing to guide the final editing and debugging process. Finally, Phase III, which is currently underway, features a feasibility trial with 10 providers and 20 families to examine the feasibility of the methodology that we propose to use in a future randomized controlled trial (RCT) as well as the feasibility and acceptability of implementing the TF-CBT e-workbook in community mental health and child welfare agencies.

Phase I: Development of the TF-CBT e-Workbook

Technical Approach to Development

The e-workbook was developed as a Web-based, rather than native (ie, device-specific) application. Although this approach requires the user to have Internet access, an advantage is that Web-based tools are accessible to providers on any of the wide range of web-connected devices. A native application would ultimately require reprogramming and updating for each operating system (Android, iOS, Windows). Although no technology can be completely "future proof," it was determined that Web-based tools would have the highest potential to remain useful in meeting the aims of this project as technology progresses. Native versions of the app will likely represent the final step toward disseminating this resource once it is fully evaluated and refined.

The application is responsive; that is, it detects the type of device that is in use and adjusts to it for optimal look and feel. For example, the application can detect an iPad and use JavaScript to convert desktop mouse actions to accelerometer actions. This functionality enables the use of features such as shaking the device to trigger a response, inertial finger drawing, and the built-in microphone. Conversely, those actions would naturally degrade to normal mouse actions if a user were to access the resource via desktop computer. As the application detects different devices, the responsive layout naturally adapts to the devices' native display parameters. This achieves a fully integrated and device-agnostic application and increases potential for adoption across practice settings.

Content Development for the TF-CBT e-Workbook

Qualitative data, collected regularly as part of consultation calls conducted in TF-CBT training programs, provided valuable direction in the technical and content development phases. Specifically, these data were used to determine which components of TF-CBT are most vulnerable to drift and what activities could be developed to overcome challenges to child engagement and provider fidelity in each TF-CBT component. For example, providers who were trained in TF-CBT reported that they used the psychoeducation, anxiety management, and coping components of TF-CBT at least 50% of the time with their child trauma cases; but that the exposure components were reported least frequently (26-50%) [40]. Therefore, greater attention was paid to emphasizing exposure-specific elements of TF-CBT rather than general psychotherapy skills. The research team also carefully reviewed data collected from expert clinicians [39] to identify areas in which technology-based resources would have high potential to enhance (1) provider fidelity; (2) child engagement in treatment activities; (3) child or caregiver understanding of key treatment concepts; (4) likelihood of skill acquisition; or (5) patient adherence to homework exercises. Taken together, these data and observations were used to create the content and format of the e-workbook activities and resources listed in Table 1.

We developed numerous resources (Table 1) or "chapters" for use by providers during individual sessions with each child. Introduction videos were created for children and are available on the first screen of each chapter. Each video depicts a teenager who explains the rationale for the chapter and presents brief examples that illustrate completion of the activity. Our decision to feature older youth (ages 15-16) in most of the videos was based on focus group feedback from children aged 8-15 years. Specifically, youth at the younger end of the age range stated that they would be equally pleased with younger or older actors in the videos, whereas youth at the older end of the age range agreed unanimously that actors should be older adolescents, and that they would likely experience very little connection to a younger actor. Some videos are brief (ie, 30-60 second) clips designed for a provider to use after introducing a concept or teaching a skill with the goal of enhancing engagement and reinforcing what the provider taught during the session (eg, videos demonstrating the CBT triangle in the cognitive coping component of TF-CBT). Some chapters feature interactive touch screen games, such as drag-and-drop activities, drawing tools, trivia-style card games, and animated relaxation activities. Each activity was developed to address an element of the TF-CBT protocol that was identified by experts and providers as challenging to implement with high fidelity and engagement.

Table 1. Patient-targeted components of the TF-CBT e-workbook by session.

Treatment concept	TF-CBT e-workbook resource	Modality
One for each component of TF-CBT[a]	Introductory videos that provide an overview for the caregiver and child about why this component of treatment is important.	Video clips featuring adolescent-aged subjects
Each TF-CBT component[a]	Interactive homework assignment checklists with activity suggestions.	Interactive application
Psychoeducation[a]	"What Do You Know?" question and answer quiz game, with "card decks" designed to facilitate child-provider education around trauma, domestic violence, sexual abuse, physical abuse, personal safety, disasters, serious accidents, and bulling/peer victimization; these decks can be personalized to each patient and provider.	Interactive touch screen activity with scorekeeping
Psychoeducation	"You are not alone" interactive graphical display that provides accurate statistics about traumatic events and emotional recovery. The provider selects a question to review with the child, and the child then estimates via touch screen interaction how many of children drawn on the screen have had experiences similar to him/her. Correct answers are given with light up figures.	Interactive touch screen activity
Psychoeducation	"Your Body" cartoon that is designed to facilitate accurate labeling of body parts via drag-and-drop touch screen activity. Both genders are represented in this activity.	Interactive drag-and-drop touch screen activity
Stress management[a]	Narrated, illustrated activity to facilitate controlled breathing exercises (eg, balloon inflating/deflating at pre-set speeds).	Interactive "game" application
Stress management	Narrated, illustrated activity to assist with progressive muscle relaxation. The user touches a muscle group on the screen, the muscle group lights up on the image, and detailed instructions are narrated as the child follows along.	Interactive application
Trauma narrative[a]	Users are presented with a drawing tool where they write and/or draw their narratives using a stylus. Handwritten text and/or illustrations are created, and can be saved or exported.	Interactive drawing application
Affective regulation[a]	This tool includes several interactive activities (eg, writing board, feelings wheel, emotions thermometer) to guide child-provider education regarding emotion identification, emotion intensity, and coping skills.	Videos and touch screen activities
Cognitive coping[a]	This chapter includes a variety of educational tools such as instructional images and video clips to guide learning and provider-child interactions. The cognitive triangle is introduced. Next, children are presented with a series of videos depicting children in a variety of ambiguous situations, and are prompted to identify and discuss with their providers about their thoughts, emotions, and behaviors.	Videos and touch screen activities
In vivo exposures	Illustrated tool that uses audio narration to guide provider-child discussion around development of an exposure hierarchy by choosing exposure activities that are safe, feasible, and relevant. Narrations and illustrations are tailored to child sex and index trauma type.	Illustrated application with audio narration
Enhancing safety[a]	Trivia-style activity to facilitate child-provider education around OK/Not OK touch, managing bullying, help seeking, problem solving skills, spotting danger-signal cues, drug refusal skills, Internet safety, and coping with ongoing stressors. These decks can be personalized to each patient and provider.	Interactive touch screen activity
Conjoint sessions[a]	Homework activities to help the child prepare for conjoint sessions.	Homework activity

[a]This resource was identified by TF-CBT trainers as a necessary component to the toolkit (Hanson et al, 2014).

Some resources included activities for providers to complete with caregivers. These consisted of an extensive collection of video clips with narration that demonstrate a wide range of effective behavior management skills, including common mistakes and how to correct them. Videos were not intended to replace provider instruction and demonstration, but to support providers' attempts to teach caregivers how to apply skills across a broad range of situations and settings. Additional resources were developed for the provider. These consisted primarily of content that assists with session preparation tasks, such as setting session goals and potential agenda items, tracking, updating assigned homework activities, and guiding their clinical decision making as families progress in TF-CBT. We also created provider tabs for most resources that provide discussion points, tips, and ideas for supplemental exercises.

Phase II: Alpha and Beta Testing

Procedure

The primary purpose of alpha testing in software and intervention resource development is to assess reactions and obtain direct input from end users regarding design, content, and functionality. These data are needed to guide improvements

to the resource. Participants were provided with tablets (ie, iPads) in either individual interviews or focus group settings to interact with a select set of resources within the TF-CBT e-workbook. Providers were given access to all activities listed in Table 1, and children were given access to a representative sample of activities. Participants were asked a series of semistructured questions administered by trained interviewers. The questions asked participants to provide reactions to each tool, ranging from reactions to the overall "look and feel" of the interface, ease of navigation, and technical problems they experience. An average of 5 minutes was dedicated to review and discuss each resource. Qualitative data collected during these sessions were audiotaped for transcription, coding, and data analysis.

Participants

Alpha testing was completed via focus groups with providers (n=22) and via focus groups or individual interviews with children (n=24). Focus group sizes were generally between 5-8 participants.

Children aged 8-11 years and 12-16 years participated in separate groups; average age was 13.0 years. Children were recruited from clinical sites (local child advocacy and mental health treatment centers) and local schools with on-site mental health services with high rates of trauma exposure. Inclusion/exclusion criteria for clinic-referred children were consistent with those used in prior RCTs with TF-CBT: children were 8-16 years old, had a history of at least one potentially traumatic event, had current clinically elevated symptoms of PTSD, and did not have active suicidal or homicidal ideations or symptoms of psychosis. Participants recruited from the schools were also 8-16 years old but not required to have a reported trauma history or a particular symptom profile. This combined clinical/nonclinical sample was used to maximize input on the look-and-feel of the TF-CBT e-workbook in addition to applicability and clarity of content to youth in the targeted age range. Sample of children was 65% female, 86% Black/African American, 14% white.

Providers were recruited from several child advocacy and mental health centers. Providers were eligible if they were fully trained in TF-CBT and carried active child trauma cases at the time of the study. There was diversity in the provider sample with respect to discipline (64% counseling, 18% social work, 18% psychology), credentials (18% doctorate, 23% MSW/LCSW/LISW/LMSW, 36% LPC, 23% other Master's degree), and years of experience delivering child trauma treatment (46% 5<years, 27% 3-5 years, 27% 1-3 years). Providers also varied in the settings in which they worked (45% mental health clinic, 55% child advocacy center), with several providers delivering TF-CBT in schools or patients' homes through outreach clinics. The provider sample was 86% female, 73% Caucasian, 18% Black/African American, 4.5% Native American, and 4.5% other (unspecified).

Data Analysis

Focus group and interview responses were audiorecorded and transcribed. First, a content analysis of the interview responses was conducted through multiple close readings of the transcriptions by two independent coders (authors on this paper). Each coder generated an independent list of thematic categories and subcategories based on their review of the data (eg, usefulness of a specific tool; age appropriateness). These themes were then further developed and ordered by the primary coder and reviewed and edited by the second coder. The coders then met in a consensus conference to discuss the categories, resolve questions, and refine the thematic categories. Once this was accomplished, the themes were again reviewed. After additional discussion to review and refine categories and resolve questions, the final thematic categories were completed and higher-order categories were developed. We have previously used similar analytic approaches to qualitative research with a range of public sector health care patient and provider populations. [39,41-43] Results of these analyses, reported in Table 2 (providers) and Table 3 (children), were used to refine and fine tune the TF-CBT e-workbook. The vast majority of recommendations given by children and providers were addressed in the context of the current grant prior to feasibility testing. Some recommendations were cost prohibitive, but can be addressed with future funding in preparation for a large-scale RCT.

Table 2. Summary of qualitative feedback from mental health providers (n=22) during alpha testing.

e-workbook component (activity)	Positive feedback (mental health providers)	Recommendations[a] and observations (mental health providers)
Psychoeducation (What Do You Know? card game)	Liked provider tab	Most providers wanted to organize their own decks
	Activity was clear	Most providers wanted to create their own unique cards
	Liked the game component	One provider asked us to label "explicit content" on cards that are particularly sensitive in nature
	Good for full age range; will increase engagement	Ability to "block" certain cards per patient
	Liked interactivity	Some providers asked for more decks of cards—like cyberbullying and Internet safety questions
Psychoeducation (You Are Not Alone activity)	Difference in race/ethnicity	Include percentages and graphical presentations for older kids
	Would use with kids 8-15 years; very helpful in teaching psychoeducation	Option to view statistics for other gender
	Liked the option to address different traumas	Update one of the statistics
		Include figures with blond and red hair
Psychoeducation (Your Body activity)	Very useful in CSA cases	For some patients, would be nice if figure emphasized mouth and hands
	Likely to engage kids	Figure is not "real-looking"
	Straightforward	
	Would be useful as a homework resource for families	
	Use with (8-10 years)	
Relaxation (breathing/PMR coaching activity)	Great resource to have for families to practice at home	Would not use with older kids (other methods like visualization)
	Use with kids 8-12 years; however, some of them noted that they would use with any age range	Balloon would likely be engaging for younger kids, not for older kids (maybe different graphic like a chest or lungs)
	Good introduction to breathing exercises	Would only use this as intro to breathing and would then practice belly breathing as usual
	Helpful in delivery of treatment	An additional imagery resource for teens could be added (pictures, relaxing sounds)
Affective regulation (Writing Board and Feelings Wheel activities)	Activities are clear	Writing board should be larger; option to type; stylus; different colors
	Feelings wheel is engaging	Feelings Wheel: Intensity scale should be more obvious; add word anchors on scale along with numbers; use faces to assist with intensity scale for younger children
	Some thought good for all ages, some thought only good for ages 8-11 years	Should have more emotions or the option to pick your own emotion
	Activities are all very helpful and engaging	
Cognitive coping (thoughts-feelings-actions activity)	Videos are great	Felt activity was too long/might lose interest
	Activity to identify T-F-A	No need for example—videos are enough
	Like the checkmark	Just have a page with CBT triangle and free-text boxes for the child to practice
	Use with any age range	

[a]Many items in the *recommendations* column already have been addressed by the development team. Recommendations made by several providers, such as the ability for providers to organize decks and create new cards in the "*What Do You Know?"* game and the recommendation to add decks addressing Internet safety and cyberbullying, were addressed by the developers in the revised e-workbook prior to the feasibility trial. Other recommendations were only voiced by one or two participants, and were considered on a case-by-case basis. Some observations made by providers related to child age; however, these observations often contradicted the perceptions of children. For example, some providers felt that two or three of

the chapters were best for children under 12 years of age; but in each of these cases older adolescents (aged 13-16 years) responded very favorably to the chapters. We have informed providers of this feedback from adolescents and have asked providers to be open-minded in the context of the feasibility trial about the value of each resource on the basis of age.

Table 3. Summary of qualitative feedback from children (n=24) during alpha testing.

e-workbook component (activity)	Positive feedback (children)	Recommendations[a] and observations (children)
Psychoeducation (What Do You Know? card game)	Using technology would help kids be more comfortable	Audio narration that reads card to you
	Most youths said that they would like the iPad-based version over the real cards	Too much white background—include more colors and enhance flexibility
		Add more colors or brighter colors
		Have themes or animation that relates to the question OR that the kid can choose to "make it their own"
Psychoeducation (You Are Not Alone activity)	N/A—Children were not asked about this activity	
Psychoeducation (Your Body activity)	N/A—Children were not asked about this activity	
Relaxation (breathing/PMR Coaching activity)	Activity makes sense	Balloon is fine, maybe integrate other graphics, like lungs, or a kid breathing in and out
	Would be helpful to learn to calm down	Change white screens to something else—have options for different background colors (preferred neon)
	App version is very straightforward	Make activity available to kids/teens at home
	Like that they can practice many times	Drawn images of children should look more like a real person
	Appropriate for full age range of youth	Have a total body video
		Have buttons in order of muscle groups
		Make girl look older
Affective regulation (Writing Board and Feelings Wheel activities)	Like the activities	Want the option to type; use different colors; make screen look like notebook paper/chalkboard
	Engaging	Have the wheel make spinning noises
	Majority would want to use tech version instead of doing these activities with paper and pencil	Do not know some words (eg, elated)
	Like the feelings of charades game	Use faces as anchors on intensity scale
		Add additional link if the kid/teen wants to learn more about something
Cognitive coping (Thoughts-Feelings-Actions activity)	N/A—Children were not asked about this activity	

[a]Many items in the *recommendations* column already have been addressed by the development team. Recommendations made by several providers, such as the ability for providers to organize decks and create new cards in the "*What Do You Know?*" game and the recommendation to add decks addressing Internet safety and cyberbullying, were addressed by the developers in the revised e-workbook prior to the feasibility trial. Other recommendations were only voiced by one or two participants, and were considered on a case-by-case basis. Some observations made by providers related to child age; however, these observations often contradicted the perceptions of children. For example, some providers felt that two or three of the chapters were best for children under 12 years of age; but in each of these cases older adolescents (aged 13-16 years) responded very favorably to the chapters. We have informed providers of this feedback from adolescents and have asked providers to be open-minded in the context of the feasibility trial about the value of each resource on the basis of age.

Beta testing followed the alpha testing phase and is a form of usability testing in which refined or revised resources are evaluated by a new set of providers for acceptability and functionality. This was to ensure that any errors existing within the TF-CBT e-workbook were identified and corrected in preparation for the feasibility trial. Members of the development and investigative team reviewed all the revised components of the resource, and additional edits and refinements to content, appearance, and functionality were made in response. Figures 1-7 include screenshots of several e-workbook activities.

Figure 1. Screenshot of the eight "decks" of virtual cards in the "What Do You Know?" psychoeducation activity. Clinicians and families select the most pertinent decks for each child. The cards within each deck are customizable by the clinician to enhance relevance to each family's needs.

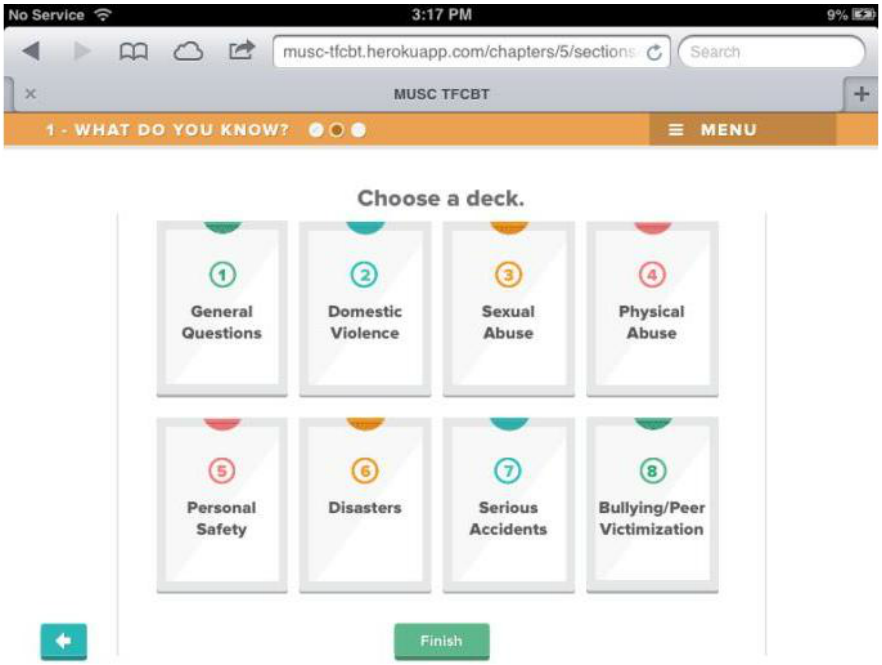


Figure 2. An example card from the Domestic Violence deck. Users can swipe left or right through the deck to view additional cards. A scoreboard is presented at the bottom to increase engagement.

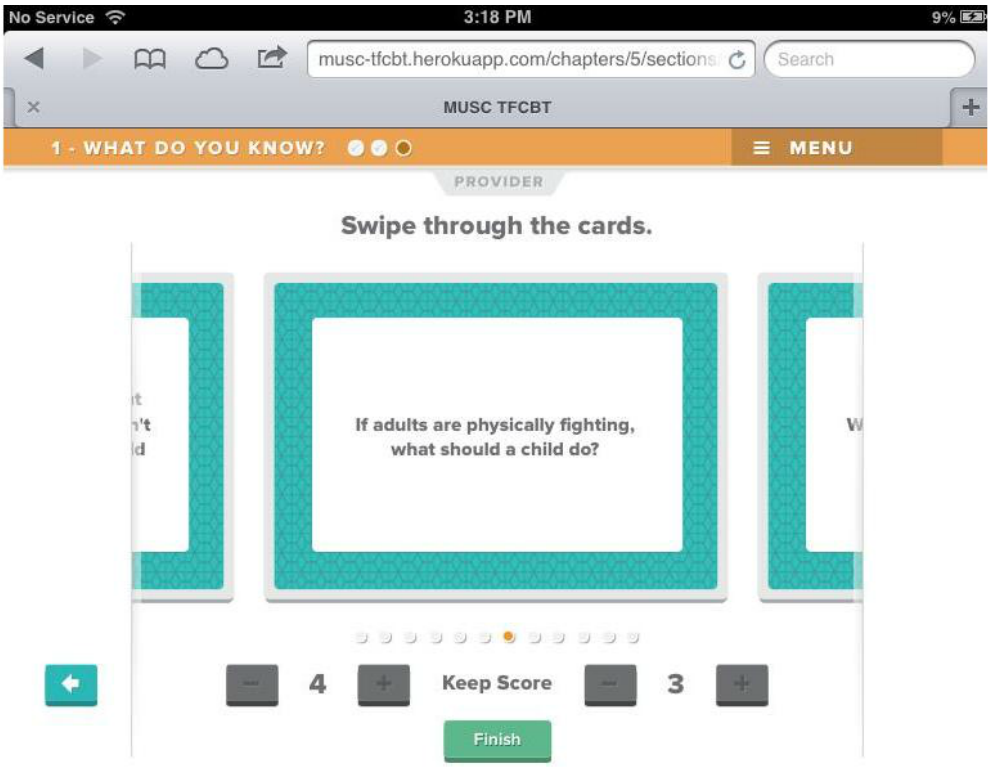


Figure 3. A Provider tab is located at the top of the screen and contains specific notes for each card. These notes may be used as discussion points during session.

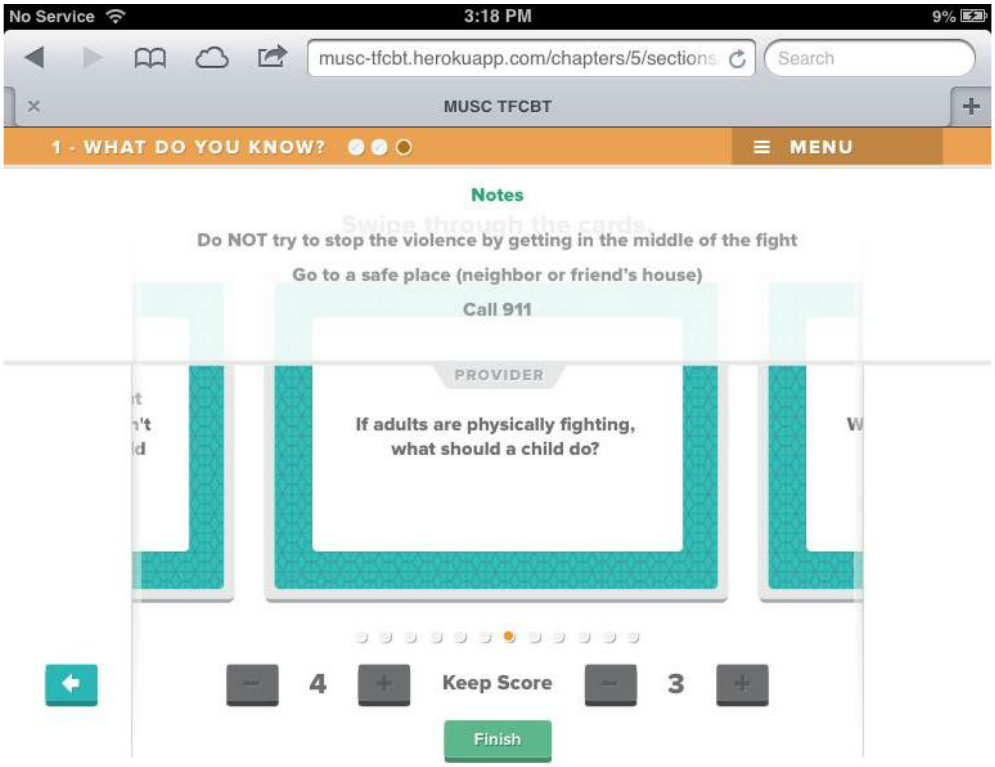


Figure 4. At the end of each activity, progress is tracked graphically via color-coding (ie, green=complete). On this page, module-specific homework assignments may be assigned. The Menu button at the top of each screen allows users to access the Home screen or end a session and assign homework.

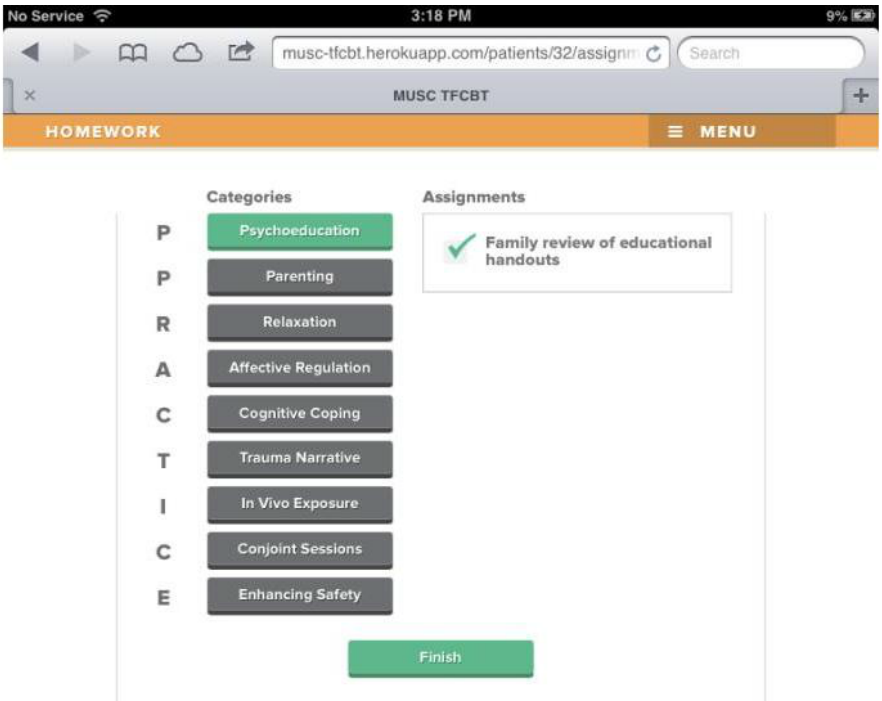


Figure 5. One activity in the Affective Regulation module involves directing the child to list as many different feeling words in 90 seconds as he or she is able to. This activity uses a free-write function. The Provider tab at the bottom of the screen provides tips for clinicians on strategies for using this activity effectively in session.

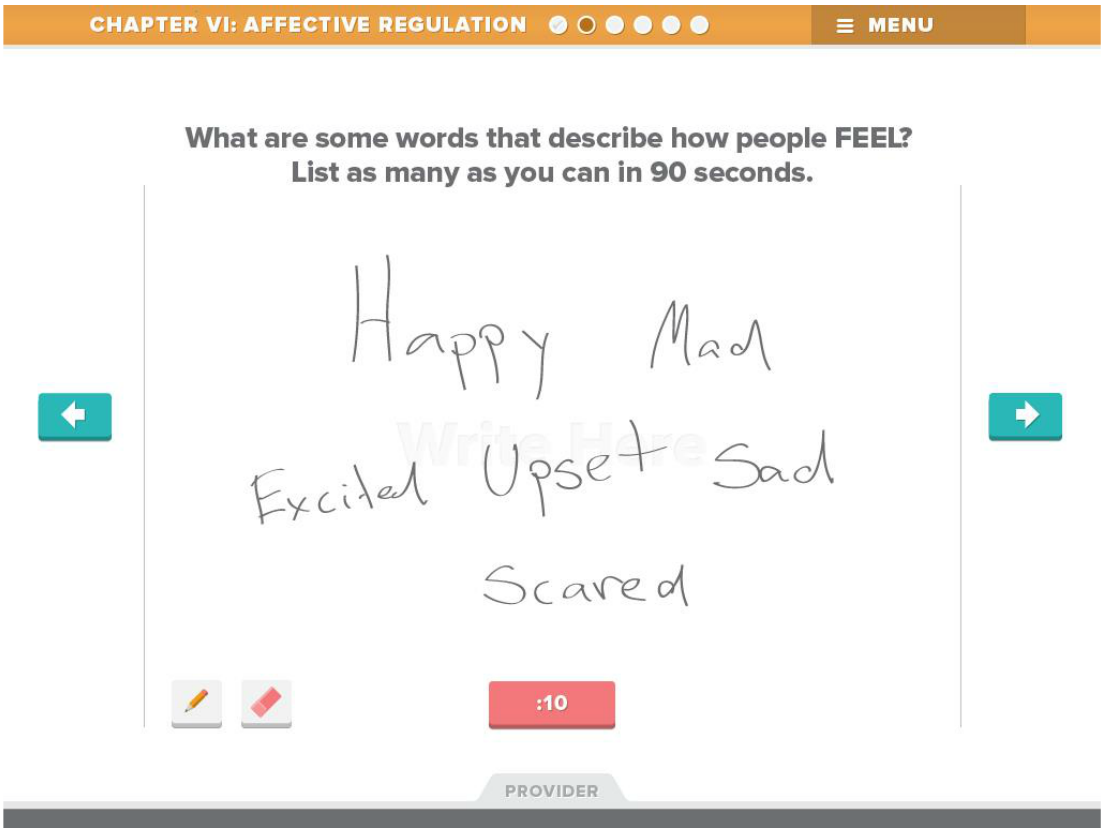


Figure 6. Another Affective Regulation activity is Feelings Charades. Users "spin" a virtual wheel by dragging down on the feeling words. They are then instructed to act out the feeling on which the wheel lands. A scoreboard is available to increase engagement with the activity.

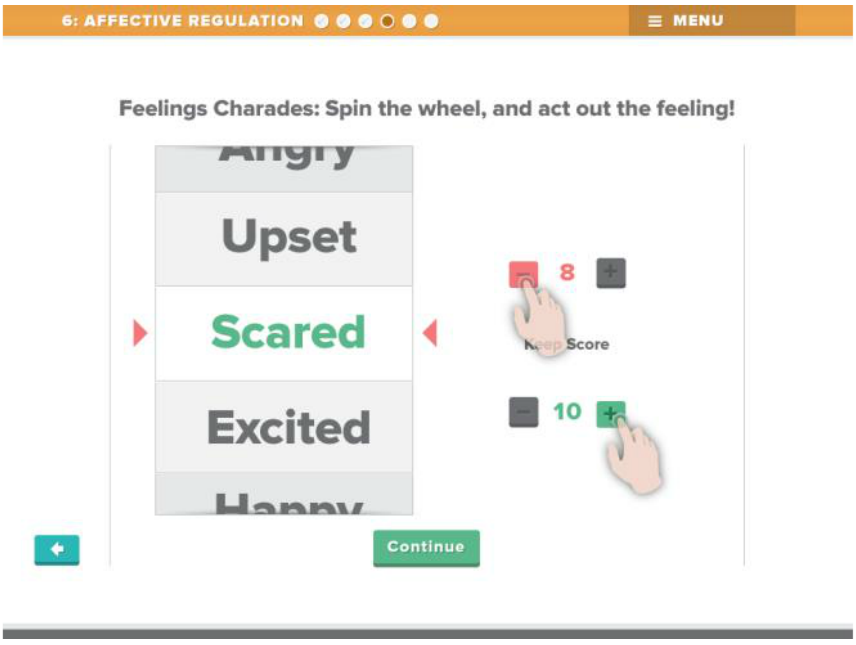
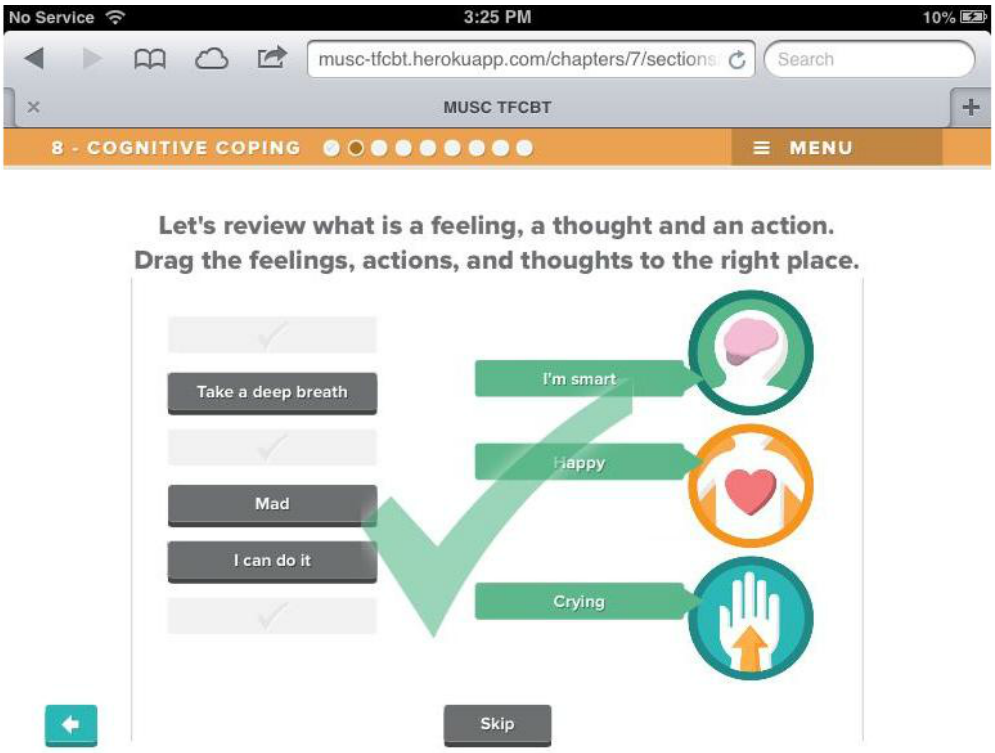


Figure 7. An activity in the Cognitive Coping module has children practice identifying and categorizing thoughts, feelings, and behaviors by dragging the words and phrases on the left of the screen (shown in gray) to the appropriate icon on the right of the screen. Feedback is provided in response to each user interaction.



Phase III: Feasibility Trial

Approach

The goal of the ongoing feasibility trial is to examine the feasibility of implementing the TF-CBT e-workbook in community mental health service agencies, and to demonstrate the randomized controlled trial (RCT) methodology we propose to use in a future large-scale trial to test the effectiveness of the TF-CBT e-workbook in clinics across the state. Resources and data resulting from this project are designed to position us well for a large-scale RCT to examine the incremental benefit of e-workbooks to best-practice interventions compared to standard care without technology. The strengths and limitations associated with conducting a small-scale feasibility RCT versus an open trial were carefully considered. A major benefit of the latter approach included increased quantity of feasibility data related to the use of the e-workbook. On balance, this option was outweighed by the importance of demonstrating the feasibility of the full RCT methodology. This would allow us to identify and address barriers in recruitment, retention, fidelity assessment, and other procedures essential to successful conduct of a RCT. This approach is consistent with expert recommendations to pilot test the feasibility of methods to be used in large RCTs, use data yielded by such studies to "debug" the methodology, and assess optimal strategies to executing the RCT [44].

Design

The feasibility trial will involve 10 providers recruited from four participating community-based mental health and child welfare agencies where TF-CBT is delivered. Providers will have been fully trained in TF-CBT and carry active child trauma cases at the onset of the trial. Providers will be block randomized by site to either the e-workbook facilitated TF-CBT (n=5) or standard TF-CBT (TAU) (n=5) conditions. Each provider will treat at least two children for a total sample of 20 children. We will also pursue an exploratory aim regarding the feasibility of using the e-workbook across multiple treatment delivery settings by including a small subsample of mental health providers who are primarily based in school settings (at least two per study condition).

An independent, trained evaluator who is blind to the study condition will conduct baseline, mid-treatment, and post-treatment clinical assessments. Families will be reimbursed $30 for each administration of the assessment battery. All sessions will be audiorecorded and coded for fidelity and child engagement by independent coders.

Alternatives to this design decision were considered carefully. For example, the design could have included each provider treating one e-workbook case and one standard TF-CBT case, with randomized ordering of conditions by provider. An advantage of this approach is that provider factors are less likely to have strong impact on clinical outcomes in this small-scale trial. A weakness of the approach is that use of the e-workbook could affect performance on subsequent standard TF-CBT cases for providers assigned to deliver TF-CBT using the e-workbook first. Additionally, it was determined that if providers within an agency were assigned to different starting conditions, this could increase risk of contamination across conditions with standard TF-CBT providers hearing about, viewing, and potentially even using elements of the e-workbook with standard

TF-CBT cases. Overall, it was concluded that block randomization by clinic to the two study conditions was a preferable design strategy.

Participants

Providers

A minimum of 10 providers who have been trained in TF-CBT and maintain active child trauma caseloads will be recruited from four participating clinics in the Charleston tri-county area. The clinics include both outpatient mental health clinics and child advocacy centers. We will make every effort to recruit a racially/ethnically diverse sample of providers to allow integration of feedback from providers with diverse backgrounds. We will also attempt to recruit a balanced sample with respect to sex and gender. Inclusion criteria for providers will be as follows. Providers will be full- or part-time employees of the participating clinic and had to have obtained at least a Master's degree in social work, counseling, clinical psychology, or a related field. Each provider will provide treatment to at least two study cases during the feasibility trial. Providers will be encouraged to enroll up to 5 children to increase the likelihood that at least 2 cases will be completed per provider during the trial.

Patients

A minimum of 20 children aged 5-15 years and their caregivers will be recruited into the feasibility trial. These children will be recruited from participating clinics in the local area. We will make every effort to recruit a racially/ethnically diverse sample of youth (eg, >25% African American) to ensure integration of feedback from patients with diverse backgrounds. Inclusion criteria will be as follows. Participating children will be aged 5-15 years, have experienced at least one potentially traumatic event (eg, sexual assault, physical assault, witnessed violence, disaster, serious accident), and have at least one symptom on each PTSD symptom cluster (re-experiencing, avoidance, hyperarousal) based on a diagnostic interview. Cases will be excluded when the child or caregiver exhibits psychotic symptoms (eg, active hallucinations, delusions, impaired thought processes); significant cognitive disability, developmental delays, or pervasive developmental disorder; or active suicidal or homicidal ideations. Children also will be excluded when there is no consistent caregiver available to participate (eg, short-term foster care placement, restrictions by child protective services). These criteria are consistent with those used in prior TF-CBT clinical trials.

Assessment

A trained evaluator both blinded to treatment condition and fully trained in the administration of all measures will administer the assessment battery. All measures are well-validated and widely used instruments in the mental health field and in the treatment outcome research. Assessment will occur on three occasions: at baseline (ie, during the screening interview), at mid-treatment (ie, after the sixth treatment session, approximately two months post-baseline), and at post-treatment (ie, after the twelfth treatment session, approximately four months post-baseline). Treatment fidelity and child engagement will be measured via observational coding of audiotapes.

Treatment Fidelity

Fidelity to the TF-CBT and e-workbook-facilitated TF-CBT protocols will be measured via coding of audiotaped treatment sessions by independent, trained raters. Treatment sessions will be audiotaped for both study conditions (n=20 x ~10 sessions average=~200 audiotapes). Ratings will be completed using a behaviorally specific coding system of TF-CBT provider behavior that was modified for the current study to ensure relevance to the e-workbook condition [45]. The coding system will be used to calculate providers' fidelity to each TF-CBT component. An additional eight items focus on general therapy skills, not specific to TF-CBT, including establishing an agenda, providing a treatment rationale, and assigning homework. Additional items were created to identify use of the e-workbook activities to differentiate the two study conditions. Two independent raters will listen to audiorecorded treatment session tapes and complete the modified fidelity measure to code the use and extensiveness of specific treatment techniques depicted on the recordings. Raters will be trained in the coding system and meet biweekly throughout the remainder of the feasibility trial to ensure maintenance of acceptable levels of accuracy and interrater reliability. Discrepant ratings will be reviewed until consensus is achieved. If the two raters cannot reach consensus, the PI and Co-Is will make final decisions.

Child Engagement

Child engagement will be measured via coding of audiotaped sessions by independent, trained raters. The Child Involvement Ratings Scale (CIRS) [46-47], a 6-item scale that measures child engagement for each session, will be used for this purpose. Four "positive" involvement items and two "negative" involvement items are rated for each session on a 6-point scale (ie, "not at all" to "a great deal present"). The positive-involvement items emphasize the extent to which children initiate discussions, demonstrate enthusiasm, self-disclose, and demonstrate understanding. Negative-involvement items address withdrawal or avoidance in treatment. Coders will provide ratings based on two 10-min segments of session audiotapes (ie, beginning at min 10 and min 40). Child engagement ratings on the CIRS have been associated with clinical outcomes [47] and provider flexibility in delivery of EBTs [48]. Excellent internal consistency and strong interrater reliability have been reported for this measure [47-48].

Child-Report of Functioning

Clinical Interview

The Kiddie Schedule for Affective Disorders and Schizophrenia for School Age Children-Present and Lifetime version (K-SADS-PL PTSD module) [49] is a semistructured interview that will be used to assess PTSD symptom levels and diagnostic status. The K-SADS-PL is well-established and used widely. It also has been used in numerous TF-CBT RCTs [33]. The K-SADS-PL also assesses functional impairment in school, social, and family life.

Self-Report Instruments

The Center for Epidemiological Studies Depression Scale for Children (CES-DC) assesses the severity of depressive symptomatology in children. The CES-DC is a 20-item

self-report depression inventory with possible scores ranging from 0-60. Scores over 15 are indicative of significant levels of depressive symptoms [50,51]. The UCLA PTSD Index for Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition (DSM-IV), Child Version (UCLA-PTSD) [52-54] will be used to assess the severity of PTSD symptomatology in children. The UCLA PTSD assesses exposure to traumatic events and all 17 DSM-IV symptoms of PTSD. Psychometric research has yielded significant support for the reliability, construct validity, and PTSD criterion-related validity [54]. The Therapeutic Alliance Scale for Children (TASC) [55-57] will be used to measure levels of therapeutic alliance. The TASC is an 8-item measure of the child's alliance with the provider using a 4-point scale. It has good internal consistency and interrater reliability [57]. The Child/Adolescent Satisfaction Questionnaire (CASQ) [58] is a 15-item instrument that assesses child satisfaction with mental health treatment.

Caregiver Report of Functioning

Clinical Interview

The K-SADS-PL [49] also will be administered to caregivers to assess children's PTSD symptoms and functional impairment.

Caregiver Report Instruments

The UCLA PTSD Index for DSM-IV, Parent Version [59] will be used to assess the severity of the child's PTSD symptomatology from the perspective of the caregiver. The Child Behavior Checklist-Parent Report (CBCL) [60] is a widely used measure of behavioral and social maladjustment in children, as perceived by the caregiver. The CBCL has strong psychometric properties. The Beck Depression Inventory (BDI) [61-62] will be used to assess severity of caregivers' depressive symptomatology. The BDI is a 21-item self-report scale of depression and is widely researched and has excellent concurrent and discriminant validity [63]. The Caregiver Satisfaction Questionnaire (CSQ) [58] is a 15-item instrument that assesses caregiver satisfaction with mental health treatment. The Working Alliance Inventory (WAI-short form) [64] is a 12-item measure of the parent-therapist alliance using a 7-point scale (ie, "never" to "always").

User Reactions to TF-CBT e-Workbook

To obtain direct input on the e-workbook's design, content, and functionality, post-treatment assessments will include semistructured interviews with children, caregivers, and providers after completion of e-workbook-facilitated TF-CBT. Patients and providers will complete semistructured interviews about their experiences with the e-workbook and how it affected treatment. Interviews will be audiotaped, transcribed, and interpreted using the same approach used during alpha testing.

Data Analysis Plan

For both conditions, several patient- and provider-level variables, as well as data collection procedures will be assessed and described. Patient-level variables will include the percentage of eligible patients recruited, treatment attrition, study retention, and session attendance. Recruitment will be assessed by the proportion of patients who agree to participate as compared to the total number solicited to enroll. Attrition will be assessed by examining the proportion of patients who prematurely terminate treatment. Qualitative analyses will be conducted to identify themes in termination. Study retention refers to the proportion of patients who complete all assessment points associated with the treatment protocol, including those who terminate treatment prematurely.

Provider-level variables will include provider recruitment to participate, fidelity to TF-CBT procedures, use of the resources within the e-workbook, and adherence to the session audiotaping protocol. Recruitment will be assessed by the proportion of providers who agreed to participate as compared to the total number approached. Provider fidelity will consist of the proportion of completed treatment components for the intervention according to the fidelity measure described previously. Providers will be interviewed at the end of treatment with e-workbook cases to provide feedback on the usability of the e-workbook activities. Other data to be summarized include kappa coefficients between independent fidelity raters, scheduling and logistical barriers to completing assessment, and communication successes and failures between study staff and clinic site staff around recruitment efforts to demonstrate feasibility of study procedures for our planned RCT.

Results

At the time of manuscript submission, Phases I and II of the study (mobile application development and refinement) were completed, and initial recruitment for Phase III (feasibility trial) is underway. No data have been cleaned or analyzed for the feasibility trial component of the project. All aspects of this federally funded study have been approved by the institutional review board (IRB) at the institution where the research is being conducted. Usability and focus group testing yielded a number of strong, favorable reactions from providers and families. Recommendations for refining the e-workbook also were provided, and these guided several improvements to the resource prior to initiating the feasibility trial, which is currently underway.

Discussion

The current protocol advances methods for developing technologies for use in mental health. First, it moves the field forward by developing and evaluating technology-based tools designed specifically to support treatment delivery and quality of care by targeting provider fidelity and, second, through engaging children. This represents a key step toward making EBTs more accessible to children and families. Existing technology-based tools largely target the (adult) patient directly by assisting them in self-care or homework adherence [25-43,63]. Such resources are not designed to support providers in the delivery of interventions with fidelity, and therefore are unlikely to have significant direct effect on the quality of care that families receive when they present for treatment in community mental health service agencies. Second, this line of research will provide valuable data about the potential for technology-based resources to enhance children's engagement in treatment. This is an important, but significantly underdeveloped, area of research that may have critical

implications for the health care field. Third, we capitalized on recent technological advances by developing Web-based applications that are optimized for mobile devices. This allows us to test the resources on a tablet (eg, iPad) while ensuring that we achieve a fully integrated and device-agnostic application that will operate with the most current technologies without the need to rewrite the application on a device-by-device basis. Fourth, we developed a wide range of tools (eg, videos, interactive games, drawing applications) that providers will use with children and adults (caregivers). This will ensure collection of valuable feasibility data that have high relevance to several design formats and target populations. Alternatively, a narrower focus on a specific patient population (eg, adults with depression) or specific type of resource (eg, videos only) would have had relatively less potential to significantly advance the field.

This study represents a unique opportunity to capitalize on the increasing use of mobile phone, Internet, and tablets by both patients and providers to support providers' efforts to deliver empirically supported treatments with a high level of fidelity. As these technologies continue to increase in popularity worldwide and within the health care field more specifically, it is essential to rigorously test the usability, feasibility, acceptability, and effectiveness of these resources. It is also essential to use input from patients and providers to drive decision-making related to development of these resources to ensure that they can be seamlessly integrated into practice. This study takes an initial step toward evaluating the feasibility and utility of implementing best-practice treatment with the assistance of a patient- and provider-centered tablet-based e-workbook resource. If feasibility is supported by our pilot trial, we will propose a rigorous, statewide efficacy evaluation powered to examine the impact on child mental health outcomes. Data yielded from such an evaluation will have tremendous value for purposes of developing and disseminating highly accessible resources that are designed to enhance the quality of treatment delivered to children and families in a wide range of service settings.

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Conflicts of Interest

None declared.

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Abbreviations

BDI: Beck Depression Inventory
CASQ: Child/Adolescent Satisfaction Questionnaire
CBLC: Child Behavior Checklist
CBT: cognitive behavioral therapy
CES-DC: Center for Epidemiological Studies Depression Scale for Children
CIRS: Child Involvement Ratings Scale
CSQ: Caregiver Satisfaction Questionnaire
DSM-IV: Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition
EBT: evidence-based treatment
e-workbook: tablet-facilitated Trauma-Focused CBT (mobile application)
IRB: institutional review board
KSADS-PL: Kiddie Schedule for Affective Disorders and Schizophrenia for School Age Children-Present and Lifetime version
PTSD: posttraumatic stress disorder
RCT: randomized controlled trial
TAC-R: Treatment Adherence Checklist-Revised
TASC: Therapeutic Alliance Scale for Children
TAU: treatment-as-usual
TF-CBT: Trauma-Focused Cognitive Behavioral Therapy
UCLA-PTSD: UCLA PTSD Index for DSM-IV
WAI: Working Alliance Inventory

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Original Paper

A "Community Fit" Community-Based Participatory Research Program for Family Health, Happiness, and Harmony: Design and Implementation

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Abstract

Background: A principal factor in maintaining positive family functioning and well-being, family communication time is decreasing in modern societies such as Hong Kong, where long working hours and indulgent use of information technology are typical.

Objective: The objective of this paper is to describe an innovative study protocol, "Happy Family Kitchen," under the project, "FAMILY: A Jockey Club Initiative for a Harmonious Society," aimed at improving family health, happiness, and harmony (3Hs) through enhancement of family communication.

Methods: This study employed the community-based participatory research (CBPR) approach, and adopted 5 principles of positive psychology and the traditional Chinese concepts of cooking and dining, as a means to connect family members to promote family health, happiness, and harmony (3Hs).

Results: In-depth collaboration took place between an academic institution and a large nongovernmental community organization association (NGO association) with 400 social service agency members. Both groups were deeply involved in the project design, implementation, and evaluation of 23 community-based interventions. From 612 families with 1419 individuals' findings, significant increases in mean communication time per week (from 153.44 to 170.31 minutes, P=.002) at 6 weeks after the intervention and mean communication scores (from 67.18 to 69.56 out of 100, P<.001) at 12 weeks after the intervention were shown. Significant enhancements were also found for mean happiness scores 12 weeks after the intervention (from 7.80 to 7.82 out of 10, P<.001), and mean health scores (from 7.70 to 7.73 out of 10, P<.001) and mean harmony scores (from 7.70 to 8.07 out of 10, P<.001) 6 weeks after the intervention.

Conclusions: This was the first CBPR study in a Hong Kong Chinese community. The results should be useful in informing collaborative intervention programs and engaging public health researchers and community social service providers, major stakeholders, and community participants in the promotion of family well-being. Furthermore, this study has generated an effective practice model for the improvement of family communication and well-being. Challenges in maintaining research rigor during data collection and program implementation were observed, and should be considered during future program planning.

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KEYWORDS

communication; community-based participatory research; family; happiness; harmony; health

Introduction

Family Communication

Communication has been identified as a core factor leading to family harmony and well-being. Inadequate communication may lead to verbal abuse, neglect, and indifferent relationships within the family, thus exerting negative effects on health, happiness, and harmony (the "3Hs") [1-2]. Effective communication—in the form of interaction through quality family time and direct verbal contact between family members—is associated with better mutual acceptance, conflict reduction, and harmonious family relationships [3].

Although Chinese individuals place great importance on family, certain changes in family structure, such as cross-border families in which parents must work outside Hong Kong, role changes such as grandparents becoming child caretakers, and excessively busy lifestyles, have led to inadequate time for and quality of family communication. [1] A recent local survey [4] showed that nearly 50% of respondents had never or only occasionally listened to and heeded their parents' advice on important matters. Similarly, over half rarely or never communicated with siblings on such matters. Our own study on family and health information trends [5] showed that half of the respondents reported a knowledge deficit in communicating positively with their family members. Therefore, interventions to improve family communication are warranted.

Community-Based Participatory Research

Community-based participatory research (CBPR) is an approach that combines both research methods and community capacity-building strategies to bridge the gap between knowledge produced through research and actual interventions and policies [6,7]. The approach is particularly attractive to academics and public health professionals struggling to address persistent problems of health care disparities in different populations [8], with the aim of combining knowledge and action for social change to improve community health and reduce inequality. CBPR is increasingly used to promote family well-being, with most studies conducted in Western populations, which have a family culture distinct from that of the Chinese.

Positive Psychology

Several studies have demonstrated the benefits of applying the principles of positive psychology to family well-being [9-12]. The principles of positive psychology are instrumental in the conceptualization of services provided to parents, family members, teachers, and other adults with children [13]. "Cooking" and "dining" form an easily achievable and well-accepted channel of communication between family members. Dining with family is valued by the Chinese as an important event, in which communication is emphasized in traditional culture, focusing on the individual's obligation to maintain family well-being. As part of the project, "FAMILY: A Jockey Club Initiative for a Harmonious Society," we combined these concepts to develop a CBPR project entitled "Happy Family Kitchen" (HFK), with a framework of 5 themes: praise and gratitude, flow, happiness, health, and savoring. The application of these elements in daily life is expected to further promote family health, happiness, and harmony (3Hs) by enhancing communication among family members more practically. Specifically, we aimed to build effective community partnerships with various stakeholders and NGOs, to establish a useful CBPR model based on positive psychology for promoting family well-being.

Objective

The main objective of this paper is to describe the project protocol entitled "Happy Family Kitchen," involving collaboration and partnership between academia and community stakeholders, using a community-based participatory approach, and having an implementation target of enhanced family functioning and communication to promote health, happiness, and harmony (3Hs). The effectiveness of a series of minimal preventive interventions in terms of structure, process, outcomes, and impact was also evaluated.

Methods

The project was planned and implemented according to the guidelines developed by Stith [14] and described as follows.

Ensure the Community is "Ready"

Community readiness is an important factor in enhancing community cooperation, participation, and resource sharing, and in maintaining the intracommunity climate and sustainability to implement prevention programs effectively [15-17]. Sufficient community capacity, recognition of existing problems, sufficient programs, and identification of key champions are essential indicators of community readiness [17-20]. Several in-depth interviews with community leaders were conducted in 2011 by the principal investigator to gain a better understanding of community needs regarding issues affecting family health, happiness, and harmony (3Hs) in Hong Kong [1]. Findings supported a need in the community to design effective education campaigns to further promote family communication to improve the 3Hs.

Develop "Community Coalitions"

Effective community coalitions facilitate good internal project functioning through an inclusive climate and mutual trust among community partners [11,13]. The HFK project collaborated with a major NGO, the Hong Kong Council of Social Service (HKCSS), which is an umbrella social service organization with more than 300 member agencies. The HKCSS was involved in recruitment, motivation of their member agencies to participate, and the design, implementation, evaluation, and dissemination of results. A steering committee including the HKCSS, governmental department, representatives of different nongovernmental organizations, and different community stakeholders was formed. Various components of project design, structure, means of implementation, outcome focus, and evaluation method were discussed throughout the project.

Confirm the Project Fits the Community

Community fit is achieved by meeting the identified needs of the community and designing appropriate interventions for targeted cultural groups. Programs that fit the community tend to be flexible, responsive, cost-effective, and culturally appropriate [21,22]. In exploring the community context, risk factors related to problems and current limitations should be explored before any intervention takes place; such community fitness information was examined prior to the start of the HFK project.

Program Fidelity

Program fidelity refers to the full extent of an intervention program being delivered as planned, and is associated with positive program outcomes [16,21,23,24]. Strategies to enhance fidelity include adequate staff training [25], setting minimum dosage and adherence levels [24], and providing ongoing supervision and feedback to involved parties [16,25]. These were adopted in our projects as follows:

Training for Program Leaders

To strengthen the program's fidelity, a 2-day "train-the-trainers" workshop was organized to equip NGO social workers with essential knowledge and skills to design and implement intervention programs for families. After this training, social workers designed their own programs focusing on 1 of 5 themes chosen by them with the same primary aim to improve family communication, using family cooking or dining as a platform. Each activity-based program consisted of 1-2 sessions of at least 2 hours each concerned with the core content, and 1 shorter booster session. Program content was designed according to principles of the chosen theme and characteristics and needs of the specific participants or users (eg, families with school-age children, elderly members, or disabled persons).

A set of requirements for the specific program was developed and monitored by the project steering committee as follows. The target population was to be not less than 50 families with at least two members per family in each activity. The program structure was to consist of 1 or 2 core intervention sessions and 1 booster session 6 weeks later. The theme was to be 1 of 5 designated themes of positive psychology, to be chosen by the social workers of each service agency. Finally, the program was to be delivered by trained social workers.

Process Evaluation

Given its importance as a component of a comprehensive assessment [7], a detailed process evaluation was conducted on our project to assess the quality and fidelity of the intervention delivered and the acceptability and feasibility of the programs, and to identify areas for improvement. Comprehensive evaluation tools were used and included a standardized program observation form, participant questionnaire evaluation, attendance records, and reports from the HKCSS and participating social service agencies.

A standardized program observation form was developed to monitor activities by 2 observers from Hong Kong University (HKU) and 2 from the HKCSS. The form assessed content fidelity, participant levels of interest, and program involvement, program interactivity level of delivery methods, strategies to enhance motivation of participants, time management, program preparation and elements, resources, success of implementation, implementation quality, degree of achievement of overall objectives, barriers and difficulties encountered during the program, and manpower allocation.

Behavioral checklists and program rundowns were used to triangulate findings on program fidelity. NGO agency workers addressed checklists by identifying methods as to how programs could deliver messages and encourage desired behaviors. The content and rundown of each program were designed by individual agencies and monitored by the HKCSS and HKU. Both behavioral checklists and rundowns were used to assess program adherence, content, and doses delivered.

Provide Adequate Resources, Training, Technical Assistance, and Evaluation

Successful prevention programs require sufficient resources, which include adequate and reliable funding, stable staffing and organization, adequate training, technical assistance, and program evaluation [26-28]. Evaluation is an important element in judging the impact of a program on the targeted community and its people [27-29]. Our project fulfilled these requirements because it is a part of the larger project "FAMILY: A Jockey Club Initiative for a Harmonious Society" (FAMILY Project) funded by the Hong Kong Jockey Club Charities Trust. Adequate and reliable funding from the FAMILY project and the HKCSS provided stable and consistent staffing. A registered social worker was recruited as the project manager for project planning, initiation, and implementation throughout the project. The HKCSS also provided sufficient administrative support and technical assistance. A detailed evaluation plan, led by the University of Hong Kong (HKU), was incorporated at an early stage, which helped to define the specific objectives and outcomes. The evaluation or assessment tools, with measureable outcomes, also served to guide the design of the interventions. The research team at HKU was also funded by the FAMILY project.

Ethical Statement

Ethical approval was granted by the Institutional Review Board (IRB) of the University of Hong Kong/Hospital Authority Hong Kong West Cluster. Written informed consent was obtained and recoded verbatim, and the procedure was approved by the IRB.

Results

Ensure the Community is "Ready"

Findings from our earlier qualitative study [1] suggested that communication and family interaction were core components contributing to the family 3Hs. Families of lower socioeconomic status (SES) had fewer financial, human, and social resources. Providing tangible support (eg, educational opportunities, sufficient childcare facilities, vocational skill training) was perceived as the most effective way to improve family well-being. Hence, the HFK focused on low SES families.

A needs assessment was conducted with the local community stakeholders prior to project implementation in the low SES district of Yuen Long. The results showed that the stakeholders recognized the problem but did not have any plan to organize related interventions. This suggested that the community was in a preplanning stage and further strategies to improve community readiness were needed before any specific prevention programs could be planned [19,30]. The findings of the needs assessments also helped to identify key local champions in family services and programs, and led us to seek their support to facilitate program adoption [23,25]. The stakeholders suggested that communication and family interaction needed to be improved for the promotion of family well-being. In addition, further public education and more community resources were needed, so that many stakeholders would be mobilized to collaborate.

The CBPR approach was adopted to collaborate with NGOs and community leaders to fill the gaps between community needs and social service provision. A positive climate of program implementation was maintained, which included the confirmation of rewards, support structures, and assistance in overcoming barriers and resistance [30]. This also helped to deepen collaboration among the main NGO and service units.

Develop "Community Coalitions"

A steering committee including the HKCSS, representatives of different nongovernmental organizations, and different community stakeholders was formed. It also involved the governmental Social Welfare Department, which provided strong support for the community coalition's leadership. Several task force and work-level meetings were organized during the project planning and implementation periods. Various components of project design, structure, means of implementation, outcome focus, and evaluation method were discussed throughout the project. The opinions of both stakeholders and academics were incorporated into the project. Strong support and assistance were provided by various stakeholders and partners throughout project implementation at both the organization and community levels. A total of 19 organizations with 23 service units joined the project, with an expanded network with other organizations and local government departments, including the local district service unit of the Social Welfare Department, schools, and a community health services provider.

Confirm the Project Fits the Community

The needs assessment identified several risk factors threatening family well-being, including community factors that contributed to inadequate communication, such as geographical barriers, long working hours, and families with cross-border relations or different generations living together. We adopted simple, acceptable, and easily practiced interventions introducing positive attitudes and appropriate communication skills among family members. The idea of incorporating the notions of "kitchen," "family cooking," and "having meals together" was also well accepted by both stakeholders and NGO social workers.

We hypothesized that the family 3Hs would be improved by increasing family communication quality and time through the intervention programs. Specifically, the project was conducted in 3 phases:

Phase 1: Gestation

This phase included planning, implementing, and evaluating a series of events and programs to increase public awareness of this project and prepare social workers as interventionists. Ideas for and details of the implementation logistics of activities were identified, discussed, and carried out. These promotion and preparation activities included a training program for social workers to equip themselves with the knowledge and skills of positive psychology and the design of community-based programs on family dining and cooking. An inauguration event was held to promote the aims of this CBPR project and arouse community stakeholders' and the public's awareness of the HFK and the family 3Hs. A series of engaging activities including game booths, family workshops, and sharing by guests were held to enhance the culture of positive family communication among participants. Exhibitions and leaflet distribution were also conducted to introduce the HFK project and the concepts of family health, happiness, and harmony (3Hs).

Phase 2: Dissemination

To promote the family 3Hs, 21 community programs were designed, delivered, and evaluated using conceptual framework theory developed based on 5 themes from positive psychology, including praise and gratitude, flow, happiness, health, and savoring. These 5 themes were strategically selected based on their associations with family well-being, as follows.

"Praise and gratitude" involved the expression of thanks and an emotional sense of appreciation. Praising the strength and goodness of family members through words or actions can increase their satisfaction level and further motivate them toward positive behavior. Being recognized by family members can help reduce negative emotion, strengthen family relationships, and encourage family members to overcome obstacles [31].

"Flow" is a concentrated personal experience involving an ecstatic psychological state, achieved through the completion of a challenge using their own strength and skills, such as being focused on cooking/dining with family; people in this state are completely focused and feel that time passes quickly. Csikszentmihalyi [32] believed that the only way to take ownership of one's life is by learning to direct energy and control attention. Enjoying a "flow" moment can help to increase the ability to face obstacles and provide a buffer against the effects of negative emotion. It also helps to improve attention, self-efficacy, and stamina.

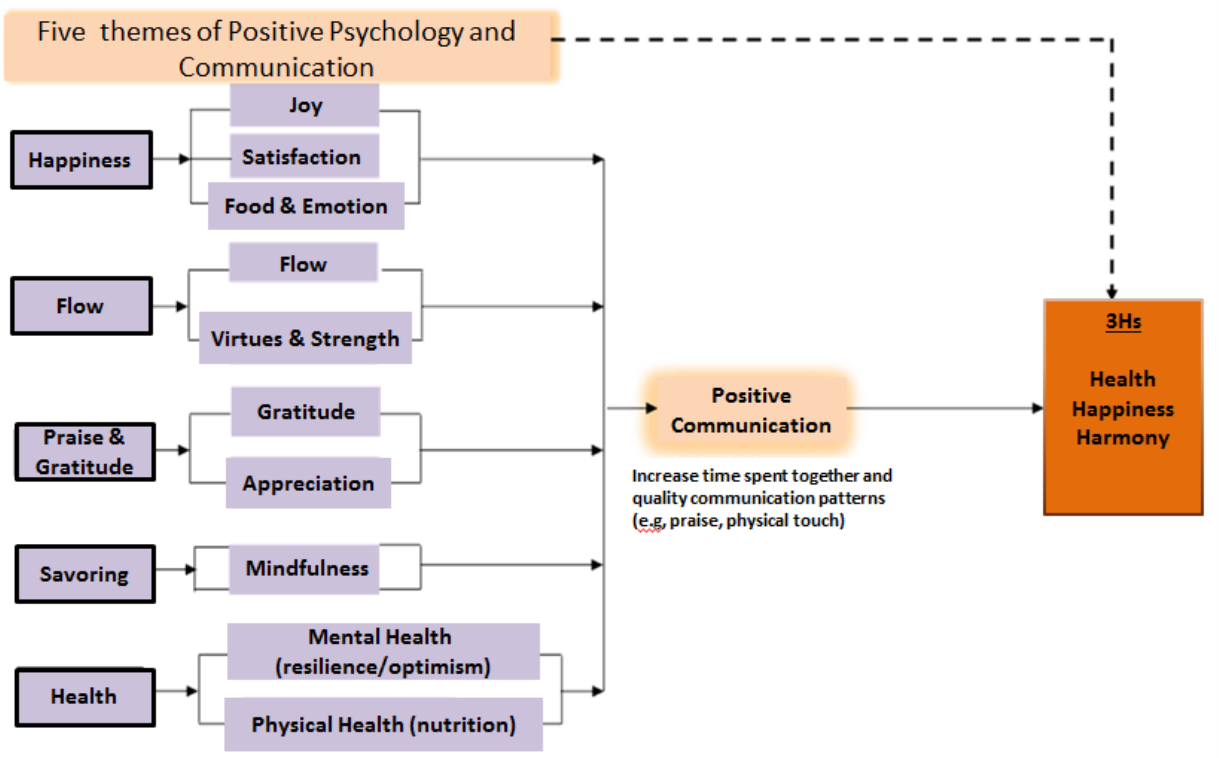
"Happiness" is a positive emotion and psychological state. It can be expressed in terms of both cognitive and affective aspects. Types of happiness include short periods of pleasure/joyfulness resulting from external stimulation without thought or consideration, and longer periods of gratification/satisfaction triggered by doing something enjoyable. Practical behavior like sharing a happy experience with family members at mealtimes improved this emotion.

"Health" can be separated into 2 aspects: mental and physical. The former is focused on the development of positive thoughts and improved resilience, and the latter on a healthy diet and better nutrition. Enjoying fruits and vegetables with family members is suggested as a simple and practical start to promoting "health" at mealtimes.

"Savoring" is not just a technique of relaxation, but also an attitude toward life: living for the present moment. It combines observing mindfulness with nonjudgmental attitudes. To practice "savoring" at mealtimes, we recommend slowing down the pace of eating and treasuring good times when dining with family. This "savoring" can help to reduce stress and depression, and broaden observation and mindset to achieve goals and become more relaxed. Langer also found that it could improve positive emotion, creativity, and physical health [33-35].

These 5 themes were integrated into a conceptual framework for our community intervention programs, as shown in Figure 1. Throughout the thematic programs, participants would experience components of one of the themes: joyfulness and satisfaction in happiness, flow and ecstasy in flow, appreciation and thankfulness in gratitude, mindfulness in savoring, and resilience or a healthy diet in health. As community-based participatory and minimal approaches were adopted, interventionists used different means of delivery; however, only 1 of the 5 theme messages was included in their programs. The aim of this design was to simplify interventions and compare the effectiveness of themes in each intervention to identify the most effective and sustainable theme. It was believed that focused thematic programs would improve the sustainability of program impact; however, because the means of message delivery vary, intervention dose may be affected. Thus, intervention hours were controlled to ensure adequate dosage in the program. Certain behavioral indicators were suggested and it was hypothesized that changes in behaviors through the 5-theme program would improve positive communication within families, by increasing their communication time or patterns, and that such enhancement would lead to the hypothesized enhanced family 3Hs. Participants' behavioral changes, family communication, and the 3Hs were evaluated both quantitatively and qualitatively.

Figure 1. Conceptual framework of the Happy Family Kitchen intervention program.



Phase 3: Consolidation

This phase included the publication and release of a "Happy Family Cookbook" to promote the project's theme and disseminate its methods and content. Finally, a practical knowledge-sharing forum was organized for social workers and other major stakeholders to showcase their programs through exhibits (including photos, feedback, and homework from participants) and discuss their experiences and the outcomes of the HFK project.

A series of territory-wide promotion and publicity strategies were implemented, including publication of the "Happy Family Cookbook" and "Practice Manual," leaflets, street banners, a website, video promotions, newspaper and magazine articles, a radio program, and the distribution of souvenirs.

Training, Proposals, and Specific Programs

Training of Social Workers

Before the intervention programs began, training sessions were delivered by a clinical psychologist and a nutritionist to introduce the theory and practice of positive psychology, and a healthy diet and nutrition, with 50 recorded attendees. HKU researchers explained scientific evaluation methods, including the principles of randomized controlled trials (RCTs). Training was interactive through the use of thematic games and group discussions. To boost the effects of this training, a set of manuals covering the content, together with proposal-writing guidelines, was distributed to participants. Moderate significant increases in perceived knowledge of positive psychology were observed immediately after training (P<.001) and were sustained at 6 months. Similar significant change was found for perceived ability to use positive psychology more frequently to advance family relationships and the 3Hs.

A total of 21 proposals from 23 units of 19 NGOs were submitted, received, revised, and vetted by the steering committee, which consisted of representatives of project organizers, academia, governmental departments, nongovernmental organizations, community stakeholders, and partners. The steering committee members provided valuable comments and suggestions regarding proposal designation and implementation and improved the intervention feasibility. Most (n=13) programs adopted gratitude as the theme; 3 adopted happiness, 2 were about health and 2 about flow, and only 1 was about savoring.

Fidelity Check

The results of a check on fidelity suggested that the community-based family programs were implemented successfully and well received by the participating families. Overall, high mean scores for program implementation (5.21, SD 1.15) and quality (5.10, SD 1.13) were rated by the observers on a Likert scale from 1 to 7. A large number of families/participants (1006 families with 2447 people) were recruited. Thematic positive psychology activities concerned with cooking and eating were attractive to many and the programs received favorable responses from almost all participants. Families demonstrated great interest in the activities, with a high level of participation, which was consistent with observers' reports (mean 5.59, SD 0.93). Furthermore, many participants showed a positive response to homework assignments. However, from the observers' view, the message delivered within programs was highly dependent on social workers' level of understanding of the selected theme and their willingness to provide high-quality services. Comprehensive questionnaires at 4 separate time points were considered too time consuming and repetitive by participants.

Evaluation

The Yuen Long district (including Tin Shui Wai) was selected because of its unfavorable social and economic environment, in which there is a high demand for community services because of low socioeconomic status and a high prevalence of domestic violence. In 2010, newly reported battered spouse cases in Yuen Long accounted for 9.6% of all cases in Hong Kong [36]. Yuen Long also had low income in 2010; the proportion of households with a monthly household income of less than HKD $10,000 was 27% in Hong Kong, while for Yuen Long it was 30.4% [37].

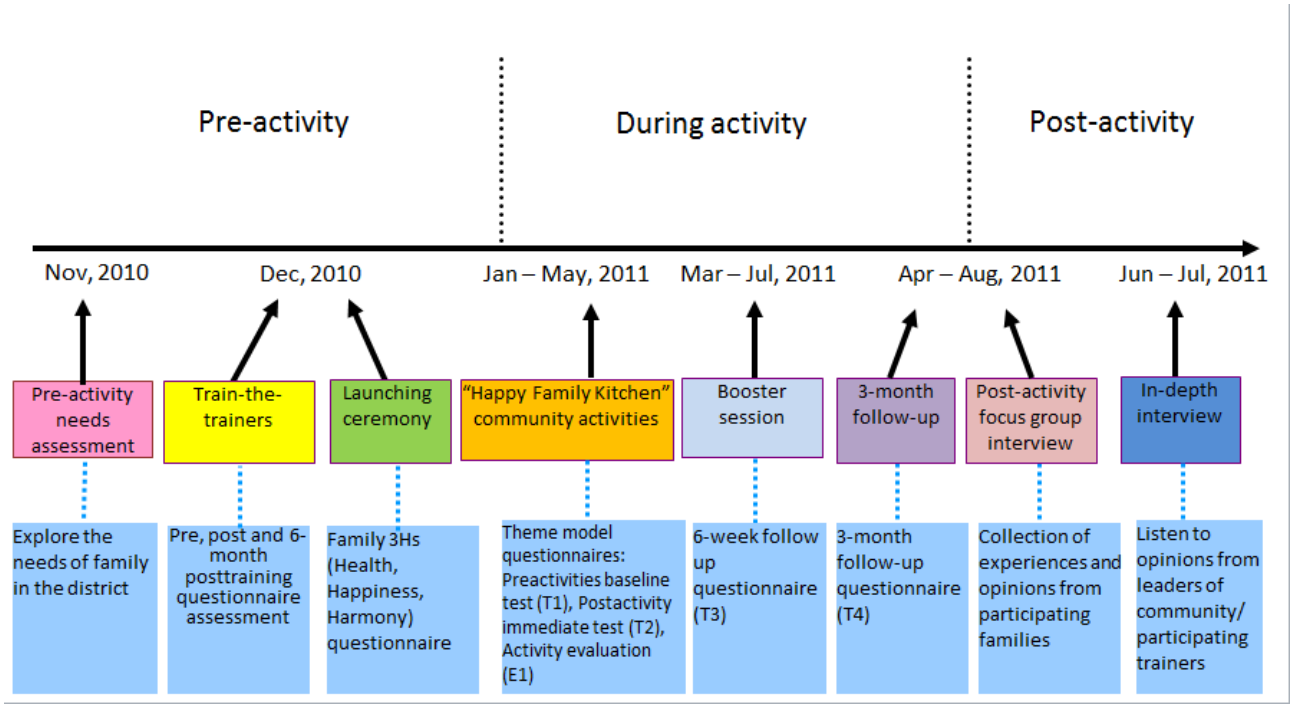
Outcome changes in both participants and social workers were measured both quantitatively and qualitatively. For participants, measures of positive psychology behavioral changes, family communication, and the 3Hs were investigated. Using qualitative methods, more in-depth information was obtained, including feelings and satisfaction levels, specific examples of behavioral changes and interpersonal relationships, and longer-term impacts on families and professionals. Details of the evaluation framework are shown in Figure 2.

The project evaluation showed improvement in key outcomes, including awareness and behaviors in positive family communication and practices, and the family 3Hs. Among 1419 individuals from 612 families, mean communication time significantly increased from 153.44 to 170.31 minutes (P=.002 per week at 6 weeks after the intervention and the mean communication score increased from 67.18 to 69.56, P<.001, out of 100 at 12 weeks after the intervention) [38]. The overall mean happiness, health, and harmony scores (range 0-10, higher scores are better) increased from 7.80 (preintervention) to 7.82 (12 weeks after the intervention; P<.001), 7.70 to 7.73 (at 6 weeks; P<.001), and 7.93 (preintervention) to 8.07 (at 6 weeks; P<.001), respectively. These findings were supported by the qualitative interviews, which showed a high level of satisfaction among participants and social workers.

Qualitative data were examined by a panel of 2 researchers; transcripts were analyzed by thematic content analysis, following the guidelines recommended by Morse and Field [39]. The results from content analyses revealed that the intervention programs, regardless of the theme applied, were effective in improving family communication. Effectiveness varied from mild to intense; for example, awareness of the importance of family communication, mutual empathy, and reflections on the parent-child relationship.

> *We would chat more while having meals together…we had never thought about that in the past…In the past, just my daughter kept talking alone…Now, my husband would start talking…actively...* [A mother, U6 16A Group 2]

Figure 2. Evaluation of the Happy Family Kitchen Project.



Discussion

Principal Findings

To the best of our knowledge, the Happy Family Kitchen project was the first CBPR in Asia to focus on promoting family communication and the 3Hs. By adopting a CBPR approach, we effectively engaged public health researchers, community service providers, major stakeholders, and local community residents in an active partnership. The project also demonstrated the feasibility of applying positive psychology in community interventions to promote the family 3Hs.

The evaluation results showed that participants generally had positive comments about the programs, in that they provided opportunities for their family members to gather and learn communication skills to improve the family 3Hs. The process evaluation also showed that the community programs were well received by participating families and appreciated by community stakeholders.

Strengths

The project adopted a CBPR approach, which could be applied to diverse social service settings and target audiences. The project fulfilled community needs: the popular topic of family cooking and dining, suggested by the community as a method to promote the family 3Hs, and the adoption of a simple intervention design, which increased project acceptability and practicality for later sustainability in the community. The fidelity results suggested that the community-based family programs were well received by participating families, who demonstrated interest in the programs and a high level of participation. Participants also were positive in their reflective feedback. The project helped to build the capacity of a cluster of local social service units to promote the family 3Hs, and the rigorous program evaluations provided clear evidence of the effectiveness of the interventions. By engaging a large number of NGOs and service units, the project may also promote the adoption of the family 3Hs through social norms in Hong Kong.

Limitations and Recommendations

Program Quality

As the interventions were developed by different NGOs, variations in the background of the NGOs, stakeholders, social workers, and other community members constituted a noticeable risk such that the dose, content delivery, and quality of the programs could vary. To increase the fidelity of program delivery, the project developed a theoretical framework to guide NGOs in design and implementation, and all program proposals were vetted by the project organizing committee to ensure they fit the theory and objectives. However, we found that a few interventionists mainly focused on positive psychology and communication without linking these to the family 3Hs.

This project was evaluated in detail, which may have created a burden for some community partners, in particular those without research experience. Future studies may consider more intensive training for such partners to build their research capacity. Communication and support should be provided between academic and community partners during the implementation process, to increase awareness of the importance of the project's goals evaluation, and improve the quality of project implementation and future project planning.

Participant Recruitment and Retention

Community NGO partners typically focused on recruiting participants through their own networks, or their "comfort zone." Although such an approach may be effective in terms of recruitment, it may limit generalizability. Many people who need the service may not know how to find it. It is important to encourage community partners to widen their "comfort zone" and proactively recruit individuals in need of these programs,

for example, those of low socioeconomic status or male participants (who are rare in CBPR programs). To extend the project's reach, different and more aggressive recruitment methods were proposed and adopted, such as door-to–door visits, street booths to communicate with the target audience, publicity work at community events, and use of the media to influence the community. However, a high attrition rate resulting from recruiting those not interested or too busy would lead to a waste of resources. Increasing communication and further explaining the project's design and setting to participants, by organizing more briefings and meetings or using phone contacts, should help to improve the retention rate.

Although the CBPR project is a new approach in program planning for service delivery, our study showed that it is feasible and has benefits for promoting the family 3Hs. The approach is fully accepted by community partners and government district social welfare offices. The low dose and simple intervention design used in our programs are applicable to regular services/delivery and can be promoted on a territory-wide scale. Thus, incorporating positive psychology into the conceptual framework of service programs deserves further investigation. Better communication among working partners and exploration of the supporting social networks would be beneficial to the implementation of future projects. Our HFK project also has implications for policy in terms of resource allocation to districts, especially for areas with individuals of low socioeconomic status.

Conclusion

In summary, this project was an innovative large-scale CBPR program, with implications for both academic and social services. Preliminary effective outcomes suggested successful implementation of the CBPR project, although this is subject to outcome analysis. This project combined the concepts of "best science" from academia and "best practice" from social services. In future programs, community workers should begin by adopting "best science" in development and evaluation, and enhancing awareness to maintain and sustain the program's impact.

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Authors' Contributions

THL, SSC, and MM conceived and designed the study and performed the experiments. CSSS, MPW, THL, KV, and SSC wrote the paper.

Conflicts of Interest

None declared.

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Abbreviations

3Hs: health, happiness, and harmony
CBPR: community-based participatory research
HFK: Happy Family Kitchen
HKCSS: Hong Kong Council of Social Service
HKU: Hong Kong University
IRB: institutional review board
NGO: nongovernmental organization
SES: socioeconomic status

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Protocol

Nutritional Risk in Major Abdominal Surgery: Protocol of a Prospective Observational Trial to Evaluate the Prognostic Value of Different Nutritional Scores in Pancreatic Surgery

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Abstract

Background: The influence of patients' preoperative nutritional status on their clinical outcome has already been proven. Therefore, patients with malnutrition are in need of additional therapeutic efforts. However, for pancreatic surgery, evidence suggesting the adequacy of existing nutritional assessment scores to estimate malnutrition associated with postoperative outcome is limited.

Objective: The aim of the observational trial "Nutritional Risk in Major Abdominal Surgery (NURIMAS) Pancreas" is to prospectively assess and analyze different nutritional assessment scores for their prognostic value on postoperative complications in patients undergoing pancreatic surgery.

Methods: All patients scheduled to receive elective pancreatic surgery at the University Hospital of Heidelberg will be screened for eligibility. Preoperatively, 12 nutritional assessment scores will be collected and patients will be assigned either at risk or not at risk for malnutrition. The postoperative course will be followed prospectively and complications according to the Clavien-Dindo classification will be recorded. The prognostic value for complications will be evaluated for every score in a univariable and multivariable analysis corrected for known risk factors in pancreatic surgery.

Results: Final data analysis is expected to be available during Spring 2016.

Conclusions: The NURIMAS Pancreas trial is a monocentric, prospective, observational trial aiming to find the most predictive clinical nutritional assessment score for postoperative complications. Using the results of this protocol as a knowledge base, it is possible to conduct nutritional risk-guided intervention trials to prevent postoperative complications in the pancreatic surgical population.

Trial Registration: germanctr.de: DRKS00006340; https://drks-neu.uniklinik-freiburg.de/drks_web/navigate.do?navigationId=trial.HTML&TRIAL_ID=DRKS00006340 (Archived by WebCite at http://www.webcitation.org/6bzXWSRYZ)

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KEYWORDS

Diagnosis Related Group system; malnutrition; nutritional assessment; nutritional score; pancreatic surgery

Introduction

Existing Evidence and Need for a Trial

Malnutrition is estimated as one of the leading causes for loss of health [1]. For hospitalized patients, the direct negative impact of malnutrition has broadly been examined [2-7]. Patients with tumorous diseases as well as patients being treated in intensive care units or in geriatric hospitals are mostly affected by negative impact of malnutrition [8-11].

To detect malnutrition, several scores have been developed. A recently published systematic review with meta-analysis investigated 32 scores with regard to their validity and predictive value for the population of hospitalized patients. The review indicated that only a small portion of scores had been fully validated and in particular, only limited scores are available for surgery. Development of new scores was considered redundant and they were not able to achieve higher sensitivity or specificity. Thus, trials investigating different scores in a specific patient population have been claimed necessary [12].

The population of surgical patients is specifically at high risk for being malnourished [13]. For some surgical indications, malnutrition has been proven as a risk of postoperative complications [14-16]. Regarding pancreatic surgery, limited data are available due to insufficient sample sizes or inhomogeneous populations. For example, the most recent pancreas-specific trial showed a correlation between the nutritional risk index and wound infections in patients after pancreaticoduodenectomy [17]. In addition, in this trial, the small sample size of 64 patients represents the major limitation.

Aim of the Trial

"Nutritional Risk in Major Abdominal Surgery (NURIMAS) Pancreas" (DRKS00006340) is a monocentric, prospective, observational trial with one study arm. The aim of this trial is to find the best suitable clinical nutritional assessment score to predict postoperative complications in patients undergoing pancreatic surgery.

Methods

Study Population

The study population will comprise adult patients undergoing pancreatic surgery at the Department of General, Visceral and Transplantation Surgery at the University Hospital of Heidelberg. All underlying diseases leading to a primary pancreatic resection will be included. Thus, the analysis will give information on a broad and representative population as seen in high-volume surgical centers (Textbox 1).

Textbox 1. Eligibility criteria.

Inclusion criteria

- Age ≥ 18 and ≤ 90 years
- Elective pancreatic surgery
- Written informed consent

Exclusion criteria

- Any former pancreatic-surgical procedures
- Language problems
- Inability to understand the trial

Diagnostic Intervention (Nutritional Assessment Scores)

Based on the most recent systematic review about existing nutritional assessment scores by Van Bokhorst-de van der Schueren [12], 11 scores have been selected that are in use in surgical patient populations [18-27]. Recently, the European Society of Clinical Nutrition and Metabolism (ESPEN) published a new consensus definition of malnutrition, the ESPEN malnutrition criteria [28]. Table 1 presents a summary of the 12 nutritional assessment scores that will be evaluated.

Table 1. Nutritional assessment scores.

Name	Classification for nutritional risk[a]
Nutritional Risk Index [18]	Normal/mild/moderate/severe
Nutritional Risk Screening Score and Revised Version [19,26]	Low/ moderate/high
Subjective Global Assessment [20]	No/ moderate/severe
Malnutrition Universal Screening Tool [21]	Low/ medium/high
Mini-Nutritional Assessment and Revised Version [22,27]	Normal/ at risk/malnourished
Short Nutritional Assessment Questionnaire [23]	Low/ moderate/severe
Imperial Nutritional Screening System I [24]	Not at risk/at risk
Imperial Nutritional Screening System II [24]	Green/amber/red
Nutritional Risk Classification [25]	Low/at risk
ESPEN Malnutrition Criteria [28]	Normal/malnourished

[a]The highest class for nutritional risk determined by the scores will be used as the study end point "at risk for malnutrition" for statistical evaluation.

Outcome Measures

The primary end point is postoperative morbidity and mortality. The most suitable score is defined as the score with the highest association of malnutrition and postoperative complication expressed as the highest lower bound of the 95% confidence interval of odds ratio.

Secondary end points are length of hospital stay, length of stay in intensive care unit, comprehensive complication index [29], place of discharge (discharge to home or discharge to rehabilitation or care facility), necessity of postoperative parenteral or enteral nutrition, and impact of malnutrition as diagnosis on hospital costs and Diagnosis Related Group (DRG) case cost.

Trial Site and Sample Size Calculation

The trial will be conducted at the Department of General, Visceral and Transplantation Surgery at the University Hospital of Heidelberg. Prevalence of malnutrition in pancreatic cancer is known to be 88% [30]. We calculated sample size with a lower prevalence of 70% for all pancreatic diseases. With a specificity and sensitivity of 95% and a confidence interval of 0.05, a total of 260 patients will be needed [31]. Patients will be consecutively recruited until the study population will consist of 260 patients with major pancreatic resections (pancreaticoduodenectomy, distal pancreatic resection, or total pancreatectomy). Based on the department's data (about 500 eligible pancreatic resections), recruitment will be completed within 12 months after inclusion of the first patient.

Planned Study Conduct and Trial Visits

All patients visiting the Department of General, Visceral and Transplantation Surgery at the University Hospital of Heidelberg and scheduled to receive elective operations will be screened. Eligible patients will be consecutively informed about the study purpose and conduct. After giving a written informed consent, patients will be questioned and examined (Visit 1) according to the investigated nutritional assessment scores (Table 2). Further, other known risk factors for postoperative complications will be noted [32,33]. If the operation is delayed for any reason, patients will be re-evaluated as long as preoperative data from questionnaires are not older than 36 hours at the time of actual operation.

Table 2. Flowchart of the NURIMAS trial-course of examinations.

Visit	1 Preoperative	2 POD 3-7	3 POD 10-14	4 Discharge or POD 30
Eligibility	X			
Informed consent	X			
Baseline data	X			
Nutritional scores	X			
Laboratory analyses	X	x	x	x
Assessment of surgical procedure		x		
Assessment of complications		x	x	x
Serious adverse events		x	x	x
Secondary end points		x	x	x

After the operation, the clinical course will be followed prospectively. Therefore, 3 planned visits will be performed. The first visit will be performed on postoperative days (PODs) 3-7, the second visit on PODs 10-14, and the last visit on the day of discharge or not later than POD 30. During these visits, complications according to Textbox 2 [34-41] will be assessed. Every postoperative complication will be rated according to the validated classification by Clavien-Dindo [42]. Further, on postoperative visits, the status of secondary end points will be evaluated.

Textbox 2. Assessed postoperative complications.

- Postoperative pancreatic fistula [34]
- Bile leak [35]
- Postpancreatectomy hemorrhage [36]
- Delayed gastric emptying [37]
- Surgical site infection [38]
- Other infections and sepsis [39]
- Chylous ascites (triglycerides in drainage) [40]
- Serious adverse event [41]

Data Management and Monitoring

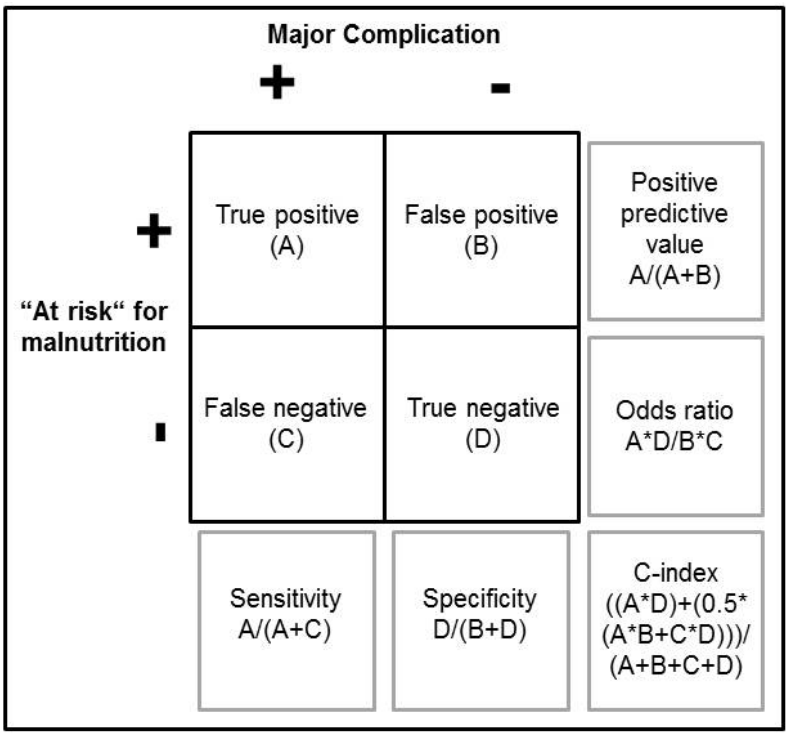
All required information according to this protocol will be recorded on a paper-based case report form. After the last visit, data will be entered in a password-protected and validated relational database (SQL Server 2008 Express). After the last patient's last visit, database will be soft-locked. A monitoring will be performed on 100% of data necessary to evaluate the primary end point. Of the remaining data, 20% are randomly selected. Finally, the database will be closed and made available for statistical analysis.

Statistical Analysis

The included scores use different numbers of nutritional risk classes. To compare the scores, evaluation of the primary end point patients will be dichotomized by each nutritional assessment score as "at risk" or "not at risk" using the respective highest nutritional risk determined by each score (Table 1). Further, patients will be dichotomized whether they had at least one major complication (Clavien-Dindo III-V) or not. Hence, for every nutritional assessment score, a contingency table will be created (Figure 1). Positive predictive value, specificity, sensitivity, and c-index will be calculated. Association between every nutritional assessment score and major complication will be expressed as odds ratio with 95% confidence interval. Univariable significance of association will be tested with a chi-square test without Yate's correction at a level of significance of 5%. A multivariable logistic regression model will be used for evaluation of primary end point at a level of significance of 5%. Covariates will be age (years) and operation time (minutes). Factors will be malignancy; gender; laparoscopy; intraoperative radiotherapy; resection of vessels (portal vein, superior mesenteric artery, or vein); inclusion in an interventional trial; American Society of Anaesthesiologists (ASA) physical status classification system; prior upper gastrointestinal surgery; pancreatic surgery associated risks (amylase in drainage >5000 IE/U on POD 1, biliary stent); and preoperative serum albumin level less than 35 g/L. Subgroup analysis will be performed separately for different types of pancreatic resections and different nutritional risk classes will be determined by each score (Table 1).

Secondary end points will be analyzed descriptively by tabulation of the measures of the empirical distributions. According to the level of the variables, means, SDs, medians, 1st and 3rd quartiles, minimum and maximum, or absolute and relative frequencies will be reported, respectively. Descriptive *P* values of the corresponding statistical tests and associated 95% confidence intervals will be given. Statistical analysis will be performed with program R [43].

Figure 1. Contingency table for calculation of primary study endpoint for the prognostic value of every nutritional assessment score.



Methods for Minimizing Bias

Minimizing Selection Bias

All patients will be consecutively screened and if found to be eligible, informed consent will be obtained in the single-arm study. Number of screened, included, and analyzed patients will be reported and differences will be explained.

Minimizing Performance and Detection Bias

Preoperative data capturing and outcome assessment will be performed by 2 different investigators. Statistical analysis will be performed after closure of database.

Minimizing Attrition Bias

Statistical measurements such as imputation will be taken to minimize risk of bias due to incomplete outcome data [44]. The trial will be reported according to the Standards for Reporting of Diagnostic Accuracy (STARD) statement [45]. The trial is registered with Deutsches Register Klinischer Studien (DRKS00006340). To avoid the risk of selective reporting, the trial protocol with full information about end points and profound explanation of planned statistical analysis is hereby published according to the Standard Protocol Items: Recommendations for Interventional Trials (SPIRIT) statement where appropriate [46]. Report on cost issues and validation of the ESPEN criteria for malnutrition is planned separately.

Minimizing Other Bias

Any financial relationship or any conflict of interest that could inappropriately influence the work within this project will be stated explicitly. Confounding will be minimized by inclusion of covariates and factors in the statistical analysis of the primary end point as mentioned in the "Statistical Analysis" section.

Ethics and Informed Consent

The NURIMAS Pancreas trial is conducted in accordance with the Declaration of Helsinki in its actual version [47]. According to the professional code for physicians in Germany (§15 BOÄ), the trial protocol was reviewed and approved by the Ethics Committee of the medical faculty of the University of Heidelberg.

Before inclusion in the NURIMAS Pancreas trial, patients will be informed both orally and in writing about all relevant aspects of the trial (eg, the aims, methods, the anticipated benefits, potential risks of the study, and the discomfort it may entail). The patients' free decision to participate will be documented by signature on the informed consent form. All patient-related information is subject to medical confidentiality and to the Federal Data Protection Act. Pseudonymized data transfer will be performed. Third parties will not have any insight into original data.

Results

Final data analysis is expected to be completed during Spring 2016.

Discussion

The NURIMAS trial is a monocentric, prospective, observational trial aiming to find the most suitable clinical nutritional assessment score to predict major postoperative complications associated with malnutrition. Thus, an important lack of knowledge in preoperative risk assessment in patients undergoing pancreatic surgery will be worked-off. Upon this knowledge, further trials can rely on a validated nutritional risk and evaluate the benefit of nutritional interventions potentially preventing postoperative complications.

Acknowledgments

This is an investigator-initiated study and PP is the primary investigator. For this study, no additional funding source is available. However, the resources and the facilities available at the University of Heidelberg are availed for conducting the trial. Only the primary investigator has access to the final dataset.

Conflicts of Interest

None declared.

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Abbreviations

ASA: American Society of Anaesthesiologists
DRG: Diagnosis Related Group
ESPEN: European Society of Clinical Nutrition and Metabolism
IMBI: Institute of Medical Biometry and Informatics
POD: postoperative day
SAE: serious adverse events
SDGC: Study Center of the German Surgical Society
SPIRIT: Standard Protocol Items: Recommendations for Interventional Trials
STARD: Standards for Reporting of Diagnostic Accuracy

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Original Paper

Exercise-Induced Tendon and Bone Injury in Recreational Runners: A Test-Retest Reliability Study

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Abstract

Background: Long-distance runners are prone to injuries including Achilles tendinopathy and medial tibial stress syndrome. We have developed an Internet comprehensive self-report questionnaire examining the medical history, injury history, and running habits of adult recreational runners.

Objective: The objective of the study was to evaluate two alternative forms of test-retest reliability of a comprehensive self-report Internet questionnaire retrospectively examining the medical history, injury history, and running habits among a sample of adult recreational runners. This will contribute to the broad aims of a wider study investigating genetics and running injury.

Methods: Invitations to complete an Internet questionnaire were sent by email to a convenience pilot population (test group 1). Inclusion criteria required participants to be a recreational runner age 18 or over, who ran over 15 km per week on a consistent basis. The survey questions addressed regular running habits and any injuries (including signs, symptoms, and diagnosis) of the lower limbs that resulted in discontinuation of running for a period of 2 consecutive weeks or more, within the last 2 years. Questions also addressed general health, age, sex, height, weight, and ethnic background. Participants were then asked to repeat the survey using the Internet platform again after 10-14 days. Following analysis of test group 1, we soft-launched the survey to a larger population (test group 2), through a local running club of 900 members via email platform. The same inclusion criteria applied, however, participants were asked to complete a repeat of the survey by telephone interview after 7-10 days. Selected key questions, important to clarify inclusion or exclusion from the wider genetics study, were selected to evaluate test-retest reliability. Reliability was quantified using the kappa coefficient for categorical data.

Results: In response to the invitation, 28 participants accessed the survey from test group 1, 23 completed the Internet survey on the first occasion, and 20 completed the Internet retest within 10-21 days. Test-retest reliability scored moderate to almost perfect (kappa=.41 to .99) for 19/19 of the key questions analyzed. Following the invitation, 122 participants accessed the survey from test group 2, 101 completed the Internet survey on the first occasion, and 50 were randomly selected and contacted by email inviting them to repeat the survey by telephone interview. There were 33 participants that consented to the telephone interview and 30 completed the questionnaire within 7-10 days. Test-retest reliability scored moderate to almost perfect for 18/19 (kappa=.41 to .99) and slight for 1/19 of the key questions analyzed. **Conclusions:** We successfully developed a self-reported, retrospective questionnaire, delivered using Internet software, providing stable and reliable answers. We demonstrate that our survey provides a relatively quick, easy to complete, and cost effective method to collect epidemiological data from recreational runners and evaluate these participants for inclusion into a genetic study.

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KEYWORDS

exercise; genetics; injury; reliability; survey

Introduction

Identification of Risk Factors for Long-Distance Runners

Several systematic reviews have examined the incidence rates and risk factors of running related injuries [1-4]. Such injuries are common, following a systematic review of the literature, Tonoli et al [4] state that injury incidence varies between 0.1% and 2.6% (P<.05). The most common injuries sustained by long-distance runners were found to be Achilles tendinopathy, iIliotibial friction syndrome, and medial tibial stress syndrome [4]. The identification of risk factors for these injury types in runners remains controversial, with conflicting results published in the literature [1,2,5-7].

There are two types of injury that are commonly reported in association with running and exercise: bone stress injuries (including medial tibial stress syndrome and tibial stress fracture) and tendon injuries (including tendinopathy and tendon rupture) [1,6]. In the case of tendon injuries, genetic risk factors have been identified that correlate with increased risk of, or protection from, injury [8]. It has been acknowledged that further investigation is required to confirm and correctly interpret the association of identified polymorphisms with specific injuries [8].

Developing the Internet Study

Published genetic studies on exercise-induced injuries primarily focus on soft tissue injuries [9], and there is only one study demonstrating association between genetic polymorphisms and increased risk of bone stress injuries [10]. In an attempt to address this, we have designed a study to analyze Australian runners using genome-wide association techniques to identify gene variants that contribute to increased risk of, or protection from, Achilles tendinopathy and/or bone stress injuries. To achieve a large sample size, the study must provide a reliable and convenient method of data collection from prospective participants, allowing them to participate in the research from home and with their own device. We therefore developed an Internet activity survey to prospectively recruit participants relevant to our test groups. Retrospective surveys of sporting injuries have been shown to have some limitations for injury epidemiology research because of issues with injury recall [11,12]. In order to counteract these issues, participants were only asked to recall injuries that had occurred in the previous 2 years.

Surveys should be evaluated for reliability, a critical measurement property for health-related and physical activity variables. Reliability refers to the consistency of answers obtained by the same participant when a measurement is repeated on different occasions [13]. Test-retest reliability is measured by having the same participants complete a survey at two different points in time to see how stable the responses are. Consistency is then quantified with a kappa coefficient statistic for categorical data [14].

Internet survey instruments are increasingly being used as a preferred method of conveniently collecting data from a wide range of participants from various locations. The reliability of survey instruments delivered on the Internet has been previously demonstrated, showing that in the instance of retrospective tobacco exposure and risk, this method of data collection is reliable [15]. Furness et al [16] also demonstrated success using an Internet survey instrument for the study of injuries collected in a retrospective manner in surfing athletes, whereby they recruited 1348 surfers. This high level of response enabled the researchers to produce a comprehensive dataset that is the largest study of surfing injuries in Australia [16].

The purpose of the study reported here is to test the test-retest reliability of an Internet survey platform from a convenience population of first responders, and pilot test participants who accepted an invitation to describe their recreational running epidemiology. This will contribute to the broad aims of the wider study, which are to recruit thousands or even tens of thousands of individuals to investigate injury epidemiology across a wide group of recreational runners in order to gain insight into how a variety of demographic factors may influence injury epidemiology; and to perform genetic assessments for the injury risk factors in a small subsample of the overall injury epidemiology study.

Methods

Self-Report Questionnaire

The questionnaire administered was designed to enable recruitment as part of the wider study entitled, “The Genetics of Exercise-Induced Injuries in Tendon and Bone.” This study has very broad aims in the range of wanting to recruit very large numbers of participants to provide injury epidemiology among recreational runners, as well as performing genetic assessments for the injury risk factors in a smaller subsample of the overall injury epidemiology study.

The platform used to deliver the survey was the Internet and commercial survey software (SurveyGizmo). The Internet survey was advertised through appropriate forums relevant to the intended population to analyze sporting injuries and training practices. Participants provided us with data on their demographics and running injuries, but the survey provides a platform for determining whether a participant met the wider genetics study’s inclusion and exclusion criteria. Participation was entirely voluntary and required consent to participate in the research processes described by checking a required “I ACCEPT” box necessary to access the questionnaire. Further information about the project was made available via a check box diverting the participant to another screen with all relevant information and contact details for further clarification if required.

On accepting conditions of participation in this project, participants were taken to the questionnaire and instructed that it would take no more than 30 minutes to complete. The software used allowed participants to exit the survey at any time and complete at a later date, allowing participants to provide their data at the time most suitable to them. A questionnaire was deemed complete when the participant had answered all required questions (customized within the software) and participants had submitted their questionnaire by checking the "SUBMIT" tab inserted at the end of the questionnaire.

The questionnaire developed here combined new items of assessment with adapted versions of the SCOFF questionnaire for examining eating disorders [17]. The full survey is provided in Multimedia Appendix 1. Regular running habits were assessed across several categories including how many years participants had been running for, how many kilometers (km) per week they ran, what terrain running was performed on, and whether orthotics were worn during running. In addition, participants reported any other sports or intentional exercise they participated in regularly over the last 2 years and were specifically asked to report any injuries of the lower limbs within the last 2 years that resulted in discontinuation of running for a consecutive period of 2 weeks or more. This was further examined by asking the participants to report on the signs and symptoms of the injury, how the injury was diagnosed, and whether it was an Achilles tendon or bone stress injury. They were also asked to stipulate any previous hip, knee, or ankle surgery over their lifetime.

In addition, participants were asked to report on their general health, provide any knowledge of existing conditions or diseases, and any known antibiotic or prescribed drug administration. They were asked to describe their regular dietary practices including sport supplement use, as well as any body weight fluctuations. Participants were also asked to report their age, sex, height, weight, and ethnic background to collect demographic data for further epidemiology analysis.

Some of the key variables, important to clarify inclusion or exclusion from the wider genetics study previously mentioned, were then selected for test-retest reliability here (Table 1). Questions of interest included those that addressed diagnosis of the injuries, as these variables may result in less than optimal reliability due to the retrospective design of the study [11]. Several questions from the tail end of the survey were also selected to assess if respondent fatigue had an effect on reliability. To approach this experimentally, we used a test-retest reliability design. This study was approved by the Human Research Ethics Committee of Bond University and the Australian Institute of Sport (RO1688B).

Table 1. Questionnaire items tested for reliability.

Questionnaire item		Possible responses
Running habits and running injuries		
	How many years have you been running on a regular basis? Regular is defined as at least weekly.	<1/1/2/3/4/5/6/7/8/9/10+
	On average, how many kilometer per week would you run?	<15/15-20/20-30/30-40/40-50/50-60/60+
	What type of terrain is the majority of your running performed on?	Bitumen/cement/hard dirt or gravel/soft dirt/grass/synthetic/treadmill
	In the last 2 years, have you participated in any other sports or intentional exercise on a regular basis (eg, weekly during at least one season)?	Yes/no
	In the last 2 years, have you had any injuries of the lower limbs, which have forced you to discontinue running for a period of 2 weeks or more?	Yes/no
	If yes, how many lower limb injuries have you been diagnosed with in the last 2 years?	1/2/3/4+
	Was the injury diagnosed by a professional doctor?	Yes/no
	Was the injury diagnosed by a professional physiotherapist?	Yes/no
	Was the injury an Achilles tendon injury?	Yes/no
	Was the injury a bone stress injury below the knee?	Yes/no
	Have you ever had hip, knee, or ankle surgery?	Yes/no
	While running do you wear orthotics?	Yes/no
General health		
	Have you ever smoked cigarettes?	Yes/no
	To your knowledge, have you ever been treated using quinolone antibiotics (eg, ciprofloxacin, norfloxacin)?	Yes/no/unsure
	To your knowledge, have you ever been treated using corticosteroid medication (eg, cortisone injection, prednisone tablets, prednisolone tablets, flixotide inhaler, pulmicort inhaler, QVAR inhaler, seretide accuhaler, symbicort turbuhaler, steroid cream)?	Yes/no/unsure
	To your knowledge, have you ever been treated using calcium tablets as prescribed by a medical doctor or taken it without a prescription?	Yes/no/unsure
	To your knowledge, have you ever been treated using vitamin D supplementation as prescribed by a medical doctor or taken it without prescription?	Yes/no/unsure
	Do you follow a gluten-free diet?	Yes/no
	Do you have any food allergies or avoidances?	Yes/no

Participants and Procedures

Test Group 1

Initial recruitment involved inviting a convenience sample of participants into a pilot group. Invitation to complete "The Genetics of Exercise-Induced Injuries in Tendon and Bone Study" survey was via email platform including an outline of the purpose of the study and providing potential participants with a link to the Internet survey. Participants were recruited from within our association or among associated colleagues. Inclusion criteria for participants included being a recreational runner aged 18 or over, who ran over 15 km/week on a consistent basis.

In response to the invitation, 28 participants accessed and consented to the conditions of the study as logged by SurveyGizmo. Of these participants, 23 completed the first survey on the first occasion, and after 10-14 days were invited by email to repeat the survey for a second time, 20 completed within 10-21 days after the date of the first completion. This group was deemed test group 1 and were subjected to the Internet-Internet test-retest method.

Test Group 2

After data from test group 1 suggested that reliability was more than moderate for 19/19 of the variables analyzed, we then sought to maximize the variability in response and strength in reliability of our questionnaire by soft launching the survey to a wider population using a different test-retest method. We invited a larger convenience sample of participants by promoting the wider genetics study through a local running club of 900 members via email platform as described earlier for group 1. The same inclusion criteria were applied and participant information was provided at the commencement of the survey, and checking the box "I ACCEPT" resulted in access to the Internet survey and provided informed consent.

In response to the invitation, 122 participants accessed and consented to the conditions of the study as logged by SurveyGizmo. Of these participants, 101 completed the first survey on the Internet on the first occasion. Of these participants, 50 were randomly selected and contacted by email platform inviting them to repeat the survey by telephone interview. In response to this invitation, 33 participants agreed to the repeat by telephone interview within 7-10 days of Internet completion and 30 answered the call within 14 days and completed the questionnaire on the second occasion. This group was deemed test group 2 and participants were subjected to the Internet-phone test-retest method.

Data Analyses

Descriptive statistics were used to describe the cohort characteristics of participants for each group, and reported as a percentage (%) of total participant number within the group illustrated. Some of the key variables important to clarify inclusion or exclusion from the wider genetics study previously mentioned were then selected for test-retest reliability and scored using the kappa statistic, with asymptotic standard error [14]. Reliability was then rated using the scale by Landis and Koch for the purposes of comparing the reliability of key questions [14]. Reliability was rated as poor (below .00), slight (.00-.20), fair (.21-.40), moderate (.41-.60), substantial (.61-.80), or almost perfect (.81-1.00). Data were collected using SurveyGizmo and analyses were conducted using SPSS Statistics version 22.0.

Results

Self-Report Questionnaire Scores

This self-report questionnaire worked toward the development of a standardized instrument via which participants not only provide us with data on their demographics and running injuries, but also provide us with a platform for determining whether interested participants meet the wider genetics study's inclusion criteria. This study analyzed the test-retest reliability of key inclusion/exclusion criteria for the wider genetics study, collected with the developed survey involving the 2 groups. The choice of different methods for test-retest analysis of each group is discussed in the "Methods" section. The results of both approaches scored using the kappa statistic, with asymptotic standard error, the valid number of responses, and number of response options, are highlighted here.

Cohort Characteristics

There were 20 participants that fulfilled group 1 criteria by completing the survey on the Internet then again on the Internet within 21 days after the first completion. Participants were between 18-68 years of age, with an equal distribution of males (50%, n=10) and females (50%, n=10). As much as 45% (9/20) of participants recorded that they had been running for 6 years or less and the remaining 55% (11/20), 8 years or more. Their running habits were variable, with 50% (10/20) predominantly running 15-30 km/week, 25% (5/20) running 30-40 km/week, and 25% (5/20) running 40 km or more per week (Table 2).

Table 2. Cohort characteristics.

Characteristics		Total participants		Test group 1 Internet-Internet		Test group 2 Internet-phone	
		N=50		N=20		N=30	
		n	%	n	%	n	%
Sex							
	Male	26	52	10	50	16	53
	Female	24	48	10	50	14	47
Age (years)							
	18-25	8	16	4	20	4	13
	26-35	19	38	8	40	11	37
	36-40	6	12	4	20	2	7
	41+	17	34	4	20	13	43
Weight (kg)							
	50-60	14	28	6	30	8	27
	61-75	24	48	9	45	15	50
	≥76	12	24	5	25	7	23
Height (cm)							
	150-160	2	4	0	0	2	7
	161-170	18	36	9	45	9	30
	171-180	16	32	5	25	11	37
	≥181	14	28	6	30	8	27
Years been running [a]							
	≤6	21	42	9	45	12	40
	≥8	29	58	11	55	18	60
Kilometers run per week [a]							
	15-30	23	46	10	50	13	43
	31-40	14	28	5	25	9	30
	≥ 41	13	26	5	25	8	27

[a]As the participants were required to describe the average distance run per week and years of running experience within certain categories, for example, 15-20 km/week, 10+ years, no means or SD could be calculated for these variables.

Participants in Group 2

There were 30 participants that fulfilled group 2 criteria by completing the survey on the Internet then again by phone interview within 14 days after Internet completion. Participants were between 18 and 67 years of age, with an almost equal distribution of males (53%, n=16) and females (47%, n=14). There were 40% (12/30) of participants that had been running for 6 years or less and the remaining 60% (18/30) 10 years or more. Their running habits were similar to test group 1 with 43% (13/30) predominantly running 15-30 km/week, 30% (9/30) 30-40 km/week, and 27% (8/30) 40 km or more per week (Table 2).

Test Group 1

Reliability was almost perfect for the type of terrain run on (kappa=.863), injuries of the lower limbs reported in the last 2 years (kappa>.99), and the number of these lower limb injuries diagnosed in the same time frame (kappa>.99). Reliability was also almost perfect for the injury being diagnosed by a doctor (kappa=.820), the injury being a bone stress injury (kappa>.99), reporting any hip/knee/ankle surgery (kappa=.857), and the use of orthotics while running (kappa>.99). Reliability was substantial for the number of years running on a regular basis (kappa=.791), average kilometer run per week (kappa=.684), other sports or intentional exercise participated in regularly in the last 2 years (kappa=.773), and reporting the injury as an Achilles tendon injury (kappa=.625). Reliability was moderate for being diagnosed by a physiotherapist (kappa=.467; Table 3).

Table 3. Test group 1 (Internet-Internet), test-retest reliability for running habits, injuries, and general health.

Test-retest reliability	n	Number of response options	Kappa (A-symp SE)
Running habits and injuries			
How many years have you been running on a regular basis? Regular is defined as at least weekly.	20	11	.791 (.100)
On average, how many kilometer per week would you run?	20	7	.684 (.119)
What type of terrain is the majority of your running performed on?	20	7	.863 (.088)
In the last 2 years, have you participated in any other sports or intentional exercise on a regular basis (eg, weekly during at least one season)?	20	2	.773 (.216)
In the last 2 years, have you had any injuries of the lower limbs, which have forced you to discontinue running for a period of 2 weeks or more?	20	2	>.99
If yes, how many lower limb injuries have you been diagnosed with in the last 2 years?	12[a]	4	>.99
Was the injury diagnosed by a professional doctor?	11[b]	2	.820 (.169)
Was the injury diagnosed by a professional physiotherapist?	8[c]	2	.467 (.323)
Was the injury an Achilles tendon injury?	12[a]	2	.625 (.333)
Was the injury a bone stress injury below the knee?	12[a]	2	>.99
Have you ever had hip, knee, or ankle surgery?	20	2	.857 (.138)
While running do you wear orthotics?	20	2	>.99
General health			
Have you ever smoked cigarettes?	19[d]	2	>.99
To your knowledge, have you ever been treated using quinolone antibiotics (eg, ciprofloxacin, norfloxacin)?	20	3	.468 (.174)
To your knowledge, have you ever been treated using corticosteroid medication?	20	3	.492 (.165)
To your knowledge, have you ever been treated using calcium tablets as prescribed by a medical doctor or taken it without a prescription?	20	3	.886 (.110)
To your knowledge, have you ever been treated using vitamin D supplementation as prescribed by a medical doctor or taken it without prescription?	20	3	>.99
Do you follow a gluten-free diet?	20	2	>.99
Do you have any food allergies or avoidances?	20	2	>.99

[a]n=20; valid=12; excluded=8

[b]n=20; valid=11; excluded=9

[c]n=20; valid=8; excluded=12

[d]n=20; valid=19; excluded=1

Reliability for General Health Questions

Reliability was almost perfect for the majority of all general health questions reported including having ever smoked cigarettes (kappa>.99), any knowledge of being treated with calcium tablets (kappa=.886), any knowledge of being treated with vitamin D supplementation (kappa>.99), reporting on whether participants followed a gluten-free diet (kappa>.99), or stating any food allergies or avoidances (kappa>.99). Reliability for any knowledge of being treated with corticosteroid medication (kappa=.492) or quinolone (kappa=.468) was moderate (Table 3).

Test Group 2

The reliability of running habit and running injury key variables in test group 1 was more variable than that of test group 1 (Table 4). Reliability was almost perfect for injuries of the lower limbs in the last 2 years, which forced discontinuation of running for a period of 2 weeks or more (kappa=.930), reporting the injury as an Achilles tendon injury (kappa=.814), reporting any hip/knee/ankle surgery (kappa>.99), and the use of orthotics while running (kappa>.99). Reliability was substantial for the number of years running on a regular basis (kappa =.639), the number of lower limb injuries diagnosed within the last 2 years (kappa=.697), injury diagnosed by a doctor (kappa=.727), and

injury diagnosed by a physiotherapist (kappa=.615). Reliability was moderate for kilometer run per week (kappa=.540), the type of terrain run on (kappa=.469), other sports or intentional exercise participated in regularly in the last 2 years (kappa=.473), and reporting the injury as bone stress injury (kappa=.571; Table 4).

Table 4. Test group 2 (Internet-phone), test-retest reliability for running habits, injuries, and general health.

Test-retest reliability	n	Number of response options	Kappa (A-symp SE)
Running habits and injuries			
How many years have you been running on a regular basis? Regular is defined as at least weekly.	30	11	.639 (.102)
On average, how many kilometer per week would you run?	30	7	.540 (.108)
What type of terrain is the majority of your running performed on?	30	7	.469 (.129)
In the last 2 years, have you participated in any other sports or intentional exercise on a regular basis (eg, weekly during at least one season)?	29[a]	2	.473 (.306)
In the last 2 years, have you had any injuries of the lower limbs, which have forced you to discontinue running for a period of 2 weeks or more?	30	2	.930 (.069)
If yes, how many lower limb injuries have you been diagnosed with in the last 2 years?	10[b]	4	.697 (.187)
Was the injury diagnosed by a professional doctor?	9[c]	2	.727 (.247)
Was the injury diagnosed by a professional physiotherapist?	10[b]	2	.615 (.337)
Was the injury an Achilles tendon injury?	11[d]	2	.814 (.175)
Was the injury a bone stress injury below the knee?	6[e]	2	.571 (.353)
Have you ever had hip, knee, or ankle surgery?	30	2	>.99
While running do you wear orthotics?	30	2	>.99
General health			
Have you ever smoked cigarettes?	30	2	.839 (.157)
To your knowledge, have you ever been treated using quinolone antibiotics (eg, ciprofloxacin, norfloxacin)?	27[f]	3	.150 (.132)
To your knowledge, have you ever been treated using corticosteroid medication?	30	3	.583 (.124)
To your knowledge, have you ever been treated using calcium tablets as prescribed by a medical doctor or taken it without a prescription?	30	3	.720 (.184)
To your knowledge, have you ever been treated using vitamin D supplementation as prescribed by a medical doctor or taken it without prescription?	30	3	.672 (.170)
Do you follow a gluten-free diet?	30	2	.783 (.209)
Do you have any food allergies or avoidances?	29[a]	2	.922 (.077)

[a]n=30; valid=29; excluded=1

[b]n=30; valid=10; excluded=20

[c]n=30; valid=9; excluded=21

[d]n=30; valid=11; excluded=19

[e]n=30; valid=6; excluded=24

[f]n=30; valid=27; excluded=3

Reliability of Test Group 1 Compared to Test Group 2

Overall, reliability was lower for general health questions reported in test group 2 in comparison to test group 1 (Table 4). Reliability was almost perfect for having ever smoked cigarettes (kappa=.839), and stating any food allergies or avoidances (kappa=.922). Reliability for any knowledge of being treated with calcium tablets (kappa=.720), any knowledge of being treated with vitamin D supplementation (kappa=.672), and reporting on whether participants followed a gluten-free diet (kappa=.783) was substantial. Reliability was moderate for any knowledge of being treated with corticosteroid medication,

however, any knowledge of being treated with quinolone had slight reliability (kappa=.150; Table 4).

Discussion

Principal Findings

The wider study, for which this survey will provide epidemiology information and a participant cohort to recruit from, will be the first, to our knowledge, to examine the genetic predisposition of tendon and bone injury in adult recreational runners. Recruitment for the larger study will involve the use of this survey, tested here initially for test-retest reliability. The results indicate that the self-reported questionnaire, delivered using the Internet commercial software, provided stable and reliable answers for many of the most important measures required for recruiting to the larger study on tendon and bone injury.

The Internet delivery of a retrospective injury study is a convenient method for remote collection of data from participants and has been used successfully in the analysis of sports-induced injuries in surfing [16] and strongman athletes [18]. As mentioned, the survey developed here combined new items of assessment with adapted versions of the SCOFF questionnaire for examining eating disorders [17]. In most aspects, its reliability was comparable to that of other similar survey delivery methods used for examining health and injury-related factors, including the Military Pre-training Questionnaire, which assesses risk factors for injury among military trainees across 5 domains (physical activity, injury history, diet, alcohol, and smoking) [19]. Test-retest reliability, for the study reported here, was shown to be acceptable for all variables for all participants, supporting the stability of the questionnaire. The only variable questionable for reliability was “treated using quinolone” (kappa=.150) for test group 2. On first completion of the Internet survey, there were 10 participants (n=30; 27 valid; 3 excluded) that were “unsure” if they had been treated in the past with quinolone. In the phone interview, these same 10 participants reported that they had not been treated previously with quinolone, and therefore this shift in reporting is responsible for the low reliability.

Poor reliability of the question addressing treatment with quinolone may suggest poor understanding of or unfamiliarity with “quinolone” upon first exposure to the question. It is known that test-retest reliability can be influenced by many factors. Interpretation of a question, such as familiarity of content or ambiguity, as well as memory can cause random answers [20]. Questions which involve unfamiliar knowledge or are ambiguous, such as asking about treatment with various prescribed medications in this survey, may lead to considerable variability in test-retest responses [21]. The nature of the question can also shift test-retest reliability scores. As we report here, it is not unusual that constant behaviors, such as smoking, result in higher reliability than more variable behavior such as obtaining an injury [11,12], or being diagnosed or treated with different medications over time [22]. Responses to questions inquiring about variable behaviors, including those addressing all aspects of running habits, would probably be more reliable if a training diary was referred to when obtaining data rather than relying on retrospective recall. However, the data presented in our study indicate moderate to almost perfect reliability for questions asking about variable behaviors without requiring participants to refer to such a tool.

Respondent fatigue is a well-known phenomenon affecting survey participants [23]. We developed this survey to minimize such fatigue by making the questions as easy to understand and answer as possible, in addition to being mindful of the time required to complete the survey. Questions regarding a gluten-free diet and known food allergies were among some of the last questions addressed in survey. We show high reliability for these variables, which supports our belief of minimal user fatigue across the questionnaire in both test groups (Tables 3 and 4). Increasing the number of possible responses did not markedly affect the reliability of the questions. Responses for questions containing only 2 variables (yes/no answers) had perfect or high reliability, while questions with 7 or 11 possible responses had moderate to high variability. This is consistent with previous research demonstrating that increasing the number of response categories only negligibly affects reliability [24,25].

Results when addressing the 2 different test groups should be considered in the context of several limitations. Our study contained a different number of participants in each group with a variable distribution of age. This limitation resulted from the sequential manner of recruitment of the two groups, inviting participation from within our networks for test group 1, then soft launching to recruit a larger group from the running club for test group 2.

There was a slightly higher test-retest reliability for test group 1, in which the survey was repeated using the same Internet platform. It is possible that this was due to learning effects, or that the retest was administered in exactly the same format.

The slightly lower test-retest reliability for group 2, in which the survey was administered by the Internet platform and then followed up by a telephone call, may be related to the wording of the question over the telephone by the interviewer compared to the participants reading the computer screen. During the telephone assessment, it is also possible that participants felt more uncomfortable answering sensitive questions or got confused in regards to what constitutes a response for each question asked. Our findings indicate that using 2 test-retest protocols resulted in participants providing the same information with high reliability for the survey administered in this study. Maintaining consistency of the protocol, however, can improve test-retest reliability.

Conclusions

We successfully developed an Internet survey to meet the recruitment needs of the larger genetic injury study. We tested its reliability using 2 different test-retest methods and used a participant sample targeted to our needs (adult recreational runners) in aid of focusing our findings to support our larger study. We had equal gender participation and a broad range of running habits and participants with reportable Achilles and bone stress injuries. Despite the aforementioned limitations, the results of this reliability study demonstrate that the Internet survey developed does provide a relatively quick, easy to

complete, and cost-effective method to collect epidemiological data from recreational runners and evaluate these participants for inclusion into a genetic study.

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Conflicts of Interest

None declared.

Multimedia Appendix 1

Survey: The genetics of exercise-induced injuries in tendon and bone.

[PDF File (Adobe PDF File), 166KB - jmir_v4i4e117_app1.pdf]

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Original Paper

Monitoring Web Site Usage of e-Bug: A Hygiene and Antibiotic Awareness Resource for Children

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Abstract

Background: e-Bug is an educational resource which teaches children and young people about microbes, hygiene, infection, and prudent antibiotic use. The e-Bug resources are available in over 22 different languages and they are used widely across the globe. The resources can be accessed from the e-Bug website.

Objective: The objective of this study was to analyze the usage of the e-Bug website in order to understand how users access the website, where and when they access the site, and to review variation in use across the different areas of the site.

Methods: The usage statistics for the e-Bug website were monitored by Google Analytics between September 2010 and August 2013.

Results: The statistics show the website had over 324,000 visits during the three years, from just under 250,000 visitors, with the number of visitors increasing year after year. Visitors accessed the website from 211 different countries, with more than 267,000 documents downloaded. The majority of visitors were from the United Kingdom and visited the English website, although countries such as France and Portugal were also frequent visitors.

Conclusions: These website statistics confirm that e-Bug is frequently used across Europe and highlight that e-Bug use has expanded across the world. The findings from this report will be used to inform future modifications or updates to the materials, as well as the development of new educational resources.

(JMIR Res Protoc 2015;4(4):e131) doi:10.2196/resprot.4049

KEYWORDS

e-Bug; educational resources; public health; antibiotics

Introduction

Controlling Antimicrobial Resistance

Antimicrobial resistance is becoming increasingly recognized worldwide as a threat to public health. A recent World Health Organization (WHO) report on antimicrobial resistance highlights the “alarming levels” of bacterial resistance seen in many parts of the world, with situations where many of the available treatment options for common infections are becoming ineffective [1]. Education of the public and professionals on the importance of controlling antimicrobial resistance through reducing antibiotic use remains a key focus of the European Centre for Disease Prevention and Control, through their annual European Antibiotic Awareness Day (EAAD) campaign, and the UK’s Department of Health, through their Five Year Antimicrobial Resistance Strategy [2]. The WHO is also urging member states to strengthen drug management systems and to support research into new treatment options [3].

The importance and effectiveness of health education websites have been demonstrated previously. A survey by the Pew Internet & American Life Project in 2009 [4] revealed that 8 out of 10 Internet users search on the Internet for health information. In addition, an analysis of the Children First for Health website revealed that 30% of inquiries regarding health topics were received directly from children and young people less than 19 years of age [5].

The Internet is a suitable tool for health promotion [6] and Internet health interventions have been shown to change behavior [7,8]. A study by Yardley et al [9] demonstrated that a Web based health intervention can lead to an increase in hand washing; suggesting hygienic behavior can be influenced through Internet tools. Evidence from the National Health Service (NHS) England annual review (2013-2014) supports that the Internet is an appropriate method for health promotion and that the Internet can be used to reach a wide target audience. The website NHS Choices has become the largest European health and care website aimed at the public, with around 27 million visitors per month [10].

The e-Bug Educational Resource

e-Bug is an educational resource for young people in the school and home setting, which aims to help reduce antimicrobial use across Europe. Children are our future generation of antibiotic users, gatekeepers, and prescribers, and education on different types of microbes and prudent antibiotic use instills these important messages at a young age. In addition, by education on key hygiene messages, such as how bacteria are spread from person to person, e-Bug aims to reduce the spread of infection through the community and hence reduce antibiotic use through this mechanism.

The e-Bug resource is led by Public Health England (formally the Health Protection Agency) and was developed in collaboration with teachers and stakeholders from 18 partner countries across Europe [11,12]. The European Directorate General funded its development for Health and Consumer Protection. Since the official launch in 2009, e-Bug has expanded across the world and, at present, has partners in 21 countries, including countries outside the European Union such as Turkey and Saudi Arabia. The resources are currently available in 22 different languages.

The e-Bug resources are hosted on the e-Bug website [13] and are all freely available for educators and students to download. The website is divided into two main sections: an area for teachers, and a separate area for students. Both areas are subdivided into junior and senior sections, for children ages 7-11 and 12-15 years, respectively. A third “Science Show” section hosts resources for children ages 5-7 years. The teacher area hosts lesson plans, activities, and worksheets covering 10 different topics, including microbes, spread of infections, antibiotics, and vaccines. The student area has Internet games and interactive activities for students to carry on learning at home.

Google Analytics has been used since 2010 to monitor Web traffic to the e-Bug website. Google Analytics has been used successfully to analyze Internet-delivered health interventions [14], and several publications have described the advantages and disadvantages to using this free service [15,16]. In addition to monitoring general website usage, changes in Web traffic can also be used to gauge the effect and success of public health awareness campaigns [17].

The aim of this report is to analyze the usage of the e-Bug website in order to understand how users access the website, where and when they access the site, and to review variation in use across the different areas of the site. This data will provide valuable information and can inform which pages should be improved and which could be promoted.

Methods

Website statistics for the e-Bug website are collected, stored, and maintained by Google Analytics.

The code for collating weblogs by Google Analytics was added into the e-Bug website from March 2010. The code allows information to be collected by the Google software, which is then filtered out by country and stored under specific country profiles created in the Google Analytics software.

For the purpose of this report, we will present results from three school academic years between September 2010 and August 2013, with each year running from September 1 to August 31. Throughout the time period covered in this report, new language websites were added to e-Bug. Weblog data for these websites were collected from the first day the pages were created.

Google Analytics was used to gather data on the number of users, number of visits, location of visitors, visit duration, bounce rate, pages visited, source of access, and downloaded files. Where number of visitors is described in this report, the figure stated is the number of unique visitors to the website and does not include returning visitors. Google Analytics defines bounce rate as the percentage of single-page sessions.

Results

General Use of the Website

Between September 1, 2010 and August 31, 2013, the e-Bug website had 249,749 users and 324,601 visits from 211 different countries across the world. The number of users has increased in each academic year (September-August) (Table 1), with a 9.64% (6866/71,237) increase in users between 2010/2011 and 2011/2012 and a 28.56% (22,308/78,103) increase in users between 2011/2012 and 2012/2013. From the total number of users across the three academic years, 76.94% (249,749/324,601) of those are new users and 23.06% (74,852/324,601) are returning users.

The bounce rate and visit duration has also improved each year since September 2010. As shown in Table 1, the bounce rate decreased every year and the visit duration for each user increased.

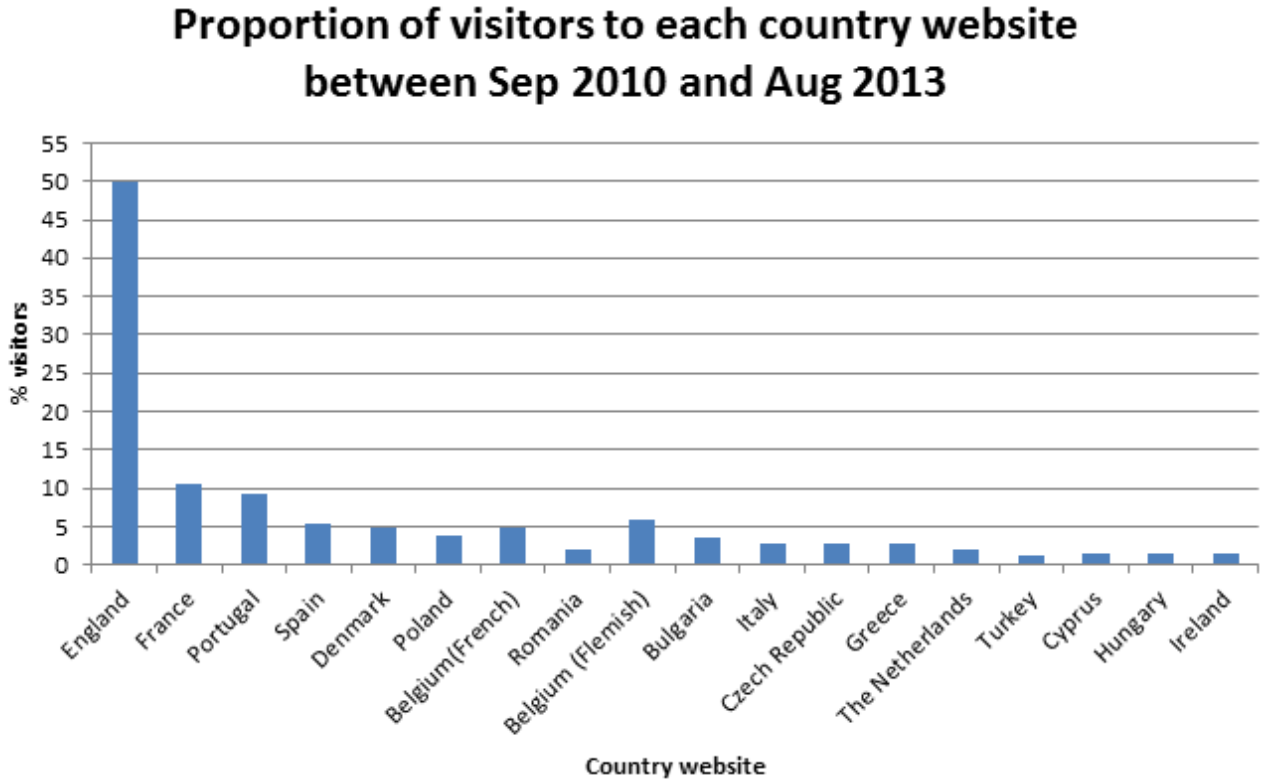
Table 1. The number, duration, and bounce rate of visitors to the e-Bug website over the last three academic years.

Year	Number of visitors	Increase in visitors from previous year, n (%)	Visit duration (minutes)	Bounce rate (%)	*P* value (increase from previous year)
2010/2011	71,237	-	4.28	36	-
2011/2012	78,103	6866/71,237(9.63)	4.51	33	.74
2012/2013	100,410	22,308/78,103 (28.56)	5.22	31	.69

General Use of the Website: Country Specific

The e-Bug website is currently available in 22 different languages, each having its own Web page which can be accessed from a menu on the home page. The English website is the most visited, with 50.45% (125,990/249,749) of all users over the three academic years, from 201 different countries, using the English site. The French and Portuguese websites are also visited frequently, with 10.53% (26,288/249,749) and 8.81% (22,013/249,749) of the users, respectively. Figure 1 shows the proportion of visitors to each country's website over the three academic years.

Figure 1. The proportion of visitors to each country's e-Bug website between September 2010 and August 2013.



General Use of the Website: Seasonal Specific

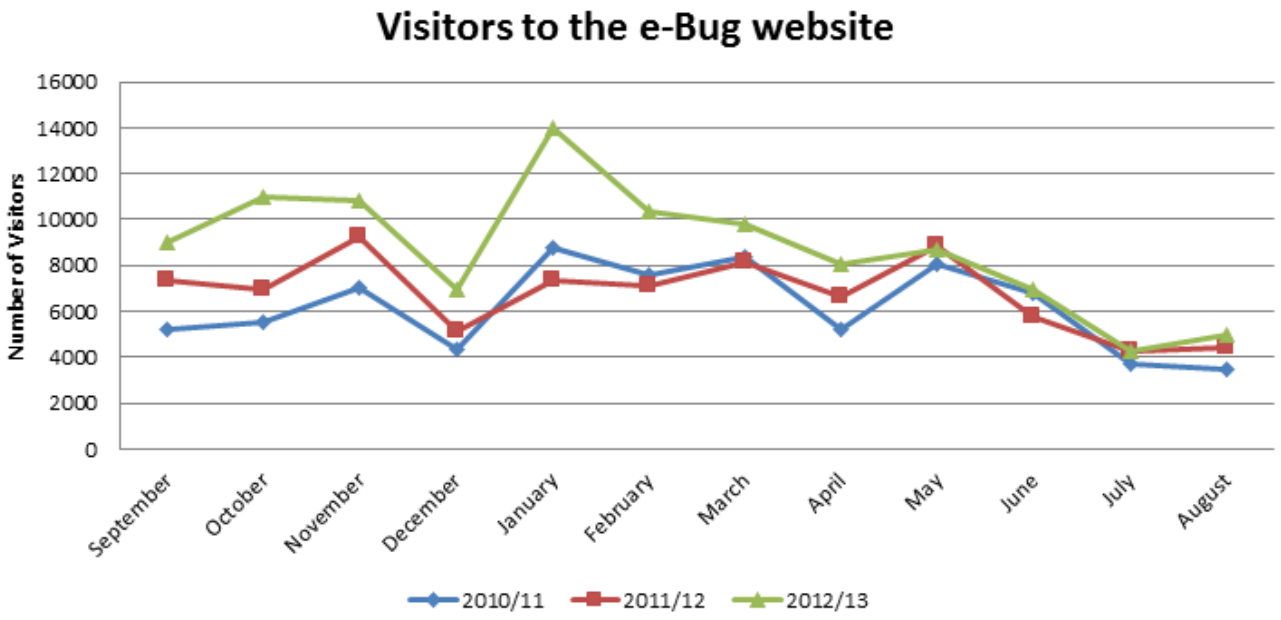
The website is accessed most during the term times of the academic years. There is always a decrease in the number of users during the Christmas (December), Easter (April), and summer holidays (July-September), and Figure 2 shows this trend clearly. In each academic year, the website gets the most visits after Christmas during January and February.

e-Bug partners across Europe often run campaigns to promote and highlight the e-Bug resources. These campaigns are particularly effective when run in line with international health campaigns, such as EAAD or Global Hand Washing Day (GHD). For example, the EAAD website hosted links to the e-Bug website during the campaign in November 2012 [18]. The e-Bug website was also hosted on the GHD website during the campaign in October 2012 [19].

For GHD in October 2012, e-Bug worked in partnership with Schools Council UK to break a Guinness World Record for the largest simultaneous hand hygiene lesson at multiple venues. The event took place across the United Kingdom, with 22 schools and 2617 participants involved. The achievement of the Guinness World Record was announced on the e-Bug website in January 2013. Between October 1, 2012 and January 31, 2013, the period during which the hand washing campaign ran, the e-Bug website had 41,643 users, which was an increase of 48.90% (13,676/27,967) from the corresponding time period the year before (27,967 users). Despite the average annual increase in users each year, the additional increase over this time period is likely due to the hand washing campaign.

The majority of visitors to the e-Bug website, 58.43% (189,653/324,601) of all visits over the three academic years, access the site through organic search engines, with Google being the most popular among users. The majority of search terms are a variation on the word “e-Bug”, such as “ebug” or “e bug”, suggesting those users are looking directly for the e-Bug website and should therefore be classed as direct visitors. Other popular search terms which bring visitors to the e-Bug website include “useful microbes” and “harmful microbes”. After organic search engines, 25.58% (83,048/324,601) of visits access the site directly through the URL, with the remaining using referral websites such as the Health Protection Agency website or the Times Educational Supplement teacher resource website.

Figure 2. The number of visitors to the e-Bug website over each academic year.



Visits From Around the World

The majority of e-Bug users are based in the United Kingdom. Over the three academic years since September 2010, visits from the United Kingdom have totaled 36.17% (117,409/324,601) of all visits, with visits from France making up 10.94% (35,502/324,601) of the total. A list of the top 10 countries visiting e-Bug can be seen in Table 2. e-Bug has partners in 24 countries who promote e-Bug to schools, however, Table 2 shows that even without an active partner, e-Bug is still used in many countries across the world. From the top 10 countries visiting e-Bug across the three academic years, there are no active partners in Brazil, United States, or India.

In total, 211 different countries accessed the e-Bug website over the last three academic years. The majority of these visits (77.52%, 251,630/324,601) came from within Europe, as shown in Table 3. After Europe, e-Bug is then used mostly in the Americas (10.58%, 34,334/324,601), followed by Asia (6.30%, 20,448/324,601), and then approximately equally in Africa and Oceania (2.40%, 7778/324,601, and 2.12%, 6886/324,601, respectively).

Analysis of the Web statistics data shows that visitors from Brazil commonly use the Portuguese e-Bug website. In the last academic year (2012/2013), there were more visits to the Portuguese website from Brazil than there were from Portugal, 3751 visits from Brazil compared with 2589 from Portugal, although this may be accounted for by the larger population in Brazil. The United States and India both access the English e-Bug website.

Table 2. Top 10 countries visiting the e-Bug website between September 2010 and August 2013.

Country	Number of visits	% of total visits, n (%)
United Kingdom	117,409	117,409/324,601 (36.17)
France	35,502	35,502/324,601 (10.94)
Portugal	15,096	15,096/324,601 (4.65)
Denmark	13,659	13,659/324,601 (4.21)
Brazil	12,303	12,303/324,601 (3.79)
Belgium	12,209	12,209/324,601 (3.76)
United States	9016	9061/324,601 (2.79)
Germany	8919	8919/324,601 (2.75)
India	8778	8778/324,601 (2.70)
Greece	8517	8517/324,601 (2.62)

Table 3. Visits to the e-Bug website from continents across the globe between September 2010 and August 2013, and split by academic year.

Continent	Number of visits (2010-2013)	% of total visits, n (%)	% of total visits split by academic year, n (%)		
			2010/2011	2011/2012	2012/2013
Europe	251,630	251,630/324,601 (77.52)	76,155/92,893 (81.98)	75,924/100,058 (75.88)	99,551/131,650 (75.62)
Americas	34,334	34,334/324,601 (10.58)	8244/92,893 (8.87)	12,357/100,058 (12.35)	13,733/131,650 (10.43)
Asia	20,448	20,448/324,601 (6.30)	4819/92,893 (5.19)	5857/100,058 (5.85)	9772/131,650 (7.42)
Africa	7778	7778/324,601 (2.40)	1925/92,893 (2.07)	2783/100,058 (2.78)	3070/131,650 (2.33)
Oceania	6886	6886/324,601 (2.12)	1443/92,893 (1.55)	2378/100,058 (2.38)	3065/131,650 (2.33)

The Most Commonly Used Resources

The e-Bug website is split into a teacher and student section, which are both further subdivided into areas for junior and senior students. Our analysis has identified that the teacher website has more users than the student website. In the 2012/2013 academic year, the most visited page in the teacher section was the “junior-e-Bug lesson pack” page, which contains the complete junior e-Bug resource pack for download, either in Word or PDF format, as well as links to each individual topic covered in the resource. This page had 19,055 visits. The most visited page in the student section was the “junior-Doctor Doctor” game with 10,820 visits.

For the English e-Bug Web pages, the junior sections in both the teacher and student areas are used more than the senior sections, with almost 4 times the number of visits to the most visited teacher pages for each section in the last year (19,055 visits compared with 5847 for senior), and 5 times more visits for the student section (10,820 visits compared with 2122 for senior). Interestingly, this trend in visits is not replicated for the French website, as here the senior website has slightly more visits than the junior website for both the teachers section and the student section. In the last year, the most visited French pages had 3259 visits for junior teacher and 4039 for senior teacher, with 566 for junior student and 633 for senior student.

The most popular topics used by teachers in all countries are "Introduction to Microbes", "Useful Microbes", "Harmful Microbes", "Food Hygiene" (for junior students only), and "Hand Hygiene". The "Farm Hygiene" resource (junior only) is used the least, most likely due to farm visits occurring mainly in the summer months, so overall usage is lower than other resources. Furthermore, not all schools visit farms and therefore fewer schools are likely to visit this page. The "Antibiotics" resource, for both junior and senior students, also has a low number of visitors.

For students, the most popular resources are the games, with "Doctor Doctor", "Chicken Surprise", and "Super Sneezes" being used the most. The "Disease Fact Files" and "Quiz" are also popular, although the "Revision Guides" and "Hall of Fame" rarely appear in the top 25 visited sections for both the junior and senior student website.

From the e-Bug Web pages, teachers can download all the resources to use within the classroom. The resources can either be downloaded as a complete pack, which contains all topics, or the individual lesson plans can be selected. Since September 2010, over 8000 complete resource packs have been downloaded from the e-Bug website, with almost 6000 of these being the junior pack. In total, there have been 267,910 downloads from the e-Bug website in the last 3 academic years. Table 4 shows these figures and identifies that almost half these downloads occurred in the 2012/2013 academic year.

Table 4. Total number of downloaded documents from the e-Bug website for each academic year.

Year	Number of downloads
2010/2011	52,673
2011/2012	73,473
2012/2013	141,774

Discussion

General Use of the Website

There has been a significant increase in the number of visitors to the e-Bug website over the last three academics years, demonstrating the need for educational resources on hygiene and infection. Between September 2010 and August 2013, the number of visitors increased by 40.95% (29,173/71,237). This increase is likely due to new international partner countries promoting e-Bug across the world, and to continued dissemination and promotion in the United Kingdom.

de Quincey et al [20] evaluated the usage of the e-Bug website between January 2008 and November 2009 using an application named Sawmill. During this time period, the website had >88,000 visitors, >169,000 downloaded documents, and an average of 3844 visitors per month. Although this raw data were cleaned before analysis with Sawmill, unlike the Google Analytics data presented here, we can still see that compared to the last year of data collection, between September 2012 and August 2013, the rate has more than doubled to an average of 8367 visitors per month.

The website statistics show that the e-Bug website is used throughout the academic year, with a decrease in the number of visitors during Christmas, Easter, and summer. This is expected as schools in Europe are closed and educators are on vacation. We can assume that users visiting the website during these holiday periods are mainly educators planning lessons for the next academic term, as the top five visited pages during these time periods are always on the teacher website. It was expected that visits to the student website would have increased during vacations, with students having more free time, however, our analysis has demonstrated that student visits also decrease during this time. It is therefore possible that student visits during term time may occur as homework related activities or as a result of being shown the website in schools.

The junior sections of the English website receive more visits than the senior sections, perhaps reflecting the number of junior versus senior schools in the United Kingdom, as of January 2014, according to the national statistics, there are five times more junior schools than senior schools [21]. This trend in visits is not seen in the French e-Bug website data, however, with the senior pages receiving slightly more visitors than the junior pages, despite there also being five times more junior schools than senior schools in France. The same pages and resources are visited most in both countries. This is likely to be due to the differences in the promotion and dissemination of e-Bug in France. The French Ministry of Education implements and promotes e-Bug in senior schools through a central mailing list; whereas junior schools are funded by their specific municipality, so they are more difficult to disseminate information to. More research is required to understand this difference in website usage.

January and February sees the most visitors to the website, which may be due to the cold and flu season, with schools wanting to highlight good hygiene practices to their students. This visit pattern is specific to England and France and is likely due to increased health promotion during the flu season and infections over the winter. The majority of overall visitors to the e-Bug website are from England and France (61.55%, 153,716/249,749), so these national campaigns will influence the overall visitors to the e-Bug website during January and February.

The Guinness World Record attempt may have contributed to the high number of visitors during January and February 2013. Visits during this time period were to all e-Bug resources, not just the hand hygiene pages, suggesting that campaigns like this encourage visits to other areas of the website. It is necessary to explore further whether e-Bug topics are generally taught in the winter term at school, in line with the National Curriculum, as this could affect the time of year that the website is visited and resources are downloaded.

Around 84.01% (272,701/324,601) of visitors to the e-Bug website access the site directly from the URL or by using search engines such as Google. The remaining 15.64% (50,756/324,601) visit the site from referral websites that link to e-Bug, suggesting that cross referencing or cross promotion of websites through similar sites is an important promotional tool.

Visits From Around the World

The e-Bug resources are used mainly in the United Kingdom, but also across Europe and the rest of world. While the number of visits from Europe has been increasing, we also see more new visitors to e-Bug from outside Europe, in particular the Americas and Asia. This confirms that e-Bug's reach is increasing across the globe. Even in countries where there has been no active dissemination of e-Bug via a partner, the website is still receiving visits. For example, the United States and India are frequently among the top 10 countries accessing the website. Having the website in languages used in more than one country has also helped increased global reach, for example, the high number of visitors in Brazil accessing the Portuguese website.

Monitoring the website statistics can give valuable information on how the website is used and the results can help inform future development and dissemination plans. The large number of visitors from Brazil suggests that there is a want by the users in that country for a health education resource of this type. Finding a partner to promote e-Bug in Brazil would ensure the resources are disseminated to as wide an audience as possible.

The Most Commonly Used Resources

It is clear from our analysis that the student website is poorly visited, with considerably fewer visits than the teacher website. To increase use, future e-Bug development may focus around new resources and tools for this group, such as games and videos. The current student resources should be further evaluated to understand what changes are necessary in order to increase use, particularly the "Revision Guides" and "Hall of Fame" resources which are used the least, only appearing once in the top 25 visited junior student pages. This lack of visitors to the "Revision Guides" is likely to be a result of the already hugely popular revision resources available to students and teachers that are linked more closely to the National Curriculum. Examples include "BBC Bitesize" and "s-cool". Promotion and dissemination of these less visited student resources should also be increased.

The most visited sections of the website are the teaching resource pages on "Introduction to Microbes", "Useful Microbes", "Harmful Microbes", "Food Hygiene", and "Hand Hygiene". These should continue to be promoted to both junior and senior schools. Poorly visited resources, such as the "Farm Hygiene" and "Antibiotics" topics, should be more widely promoted. Previous research has shown that antibiotics are not widely included in the National Curriculum of many countries, unlike other e-Bug topics, which may explain their lower use [22]. This also suggests that the curriculum has a strong influence on how the e-Bug website is used.

e-Bug is mostly used in the United Kingdom and English-speaking countries, with 36.17% (117,409/324,601) of visitors based in the United Kingdom and over 50.45% (125,990/249,749) of visitors using the English website. Dissemination of the resources across the globe should increase in correlation with the increase in active partners and the increase in languages that e-Bug resources are available in. With 22 different language websites currently live, and more under translation, it is likely that more countries will be able to use and disseminate the resources to a wider audience.

Limitations

Despite the usefulness of the Google Analytics data in tracking the use of e-Bug and visits to the website, there are also limitations. In order to fully interpret the results presented here, the data should be combined with qualitative data to understand users reasoning's and behaviors. For example, the website statistics have shown that in the United Kingdom, the junior websites are visited more than the senior websites, and the teacher websites are visited more than the students websites. Qualitative data could provide an explanation as to this difference and help guide future development or adaptations to the resources.

In addition, it is hard to gain an accurate usage of the e-Bug resources from this data as the resources are downloadable, meaning that teachers can continue to use the resources without revisiting the website. The complete e-Bug teaching pack can also be downloaded, meaning each individual topic page does not need to be visited. In 2010, printed e-Bug teaching packs were also distributed to all schools in England, meaning that teachers had no need to visit the website. It is likely that usage of e-Bug is much higher than that which is predicted from the Google Analytics data. It would be useful to explore the usage of the printed e-Bug resources distributed to all schools in England, as this may have influenced the number of visitors to the e-Bug site in that same year. It would be recommended to use qualitative research to gather this information to determine whether receiving the e-Bug teaching packs prevented teachers from visiting the website.

While the number of visits to the website is an important measurement tool of the growth of e-Bug, it is also necessary to explore the outcome of the visit to the website. Qualitative research should be conducted to determine whether the visit, and subsequent teaching, resulted in outcomes such as improved knowledge, improved enthusiasm for the subject, change in student health behaviors, for example, hand washing techniques and change in teaching methods including health topic delivery.

Implications

Results from the weblog analysis have found that qualitative work is needed to explore reasons for the poor use of the teacher "Farm Hygiene" and "Antibiotic" resources, and the student "Hall of Fame" resource and "Revision Guides". Further promotion of these resources is needed. In promotional materials to schools and educators, e-Bug should continue to highlight the most popular resources such as the "Introduction to Microbes", "Useful Microbes", and "Harmful Microbes" topics. In addition, e-Bug needs to extend partnerships to other countries speaking the languages that are available. Finally, e-Bug should be promoted via links from other health and

educational websites and other social media websites used by teachers and children.

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Conflicts of Interest

None declared.

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Abbreviations

EAAD: European Antibiotic Awareness Day
GHD: Global Hand Washing Day
NHS: National Health Service
WHO: World Health Organization

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Original Paper

Twitter-Delivered Behavioral Weight-Loss Interventions: A Pilot Series

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Abstract

Background: Lifestyle interventions are efficacious at reducing risk for diabetes and cardiovascular disease but have not had a significant public health impact given high cost and patient and provider burden.

Objective: Online social networks may reduce the burden of lifestyle interventions to the extent that they displace in-person visits and may enhance opportunities for social support for weight loss.

Methods: We conducted an iterative series of pilot studies to evaluate the feasibility and acceptability of using online social networks to deliver a lifestyle intervention.

Results: In Study 1 (n=10), obese participants with depression received lifestyle counseling via 12 weekly group visits and a private group formed using the online social network, Twitter. Mean weight loss was 2.3 pounds (SD 7.7; range -19.2 to 8.2) or 1.2% (SD 3.6) of baseline weight. A total of 67% (6/9) of participants completing exit interviews found the support of the Twitter group at least somewhat useful. In Study 2 (n=11), participants were not depressed and were required to be regular users of social media. Participants lost, on average, 5.6 pounds (SD 6.3; range -15 to 0) or 3.0% (SD 3.4) of baseline weight, and 100% (9/9) completing exit interviews found the support of the Twitter group at least somewhat useful. To explore the feasibility of eliminating in-person visits, in Study 3 (n=12), we delivered a 12-week lifestyle intervention almost entirely via Twitter by limiting the number of group visits to one, while using the same inclusion criteria as that used in Study 2. Participants lost, on average, 5.4 pounds (SD 6.4; range -14.2 to 3.9) or 3.0% (SD 3.1) of baseline weight, and 90% (9/10) completing exit interviews found the support of the Twitter group at least somewhat useful. Findings revealed that a private Twitter weight-loss group was both feasible and acceptable for many patients, particularly among regular users of social media.

Conclusions: Future research should evaluate the efficacy and cost-effectiveness of online social network-delivered lifestyle interventions relative to traditional modalities.

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KEYWORDS

social networks; Twitter; obesity; weight loss; online social networking; peer-to-peer health care; digital health

Introduction

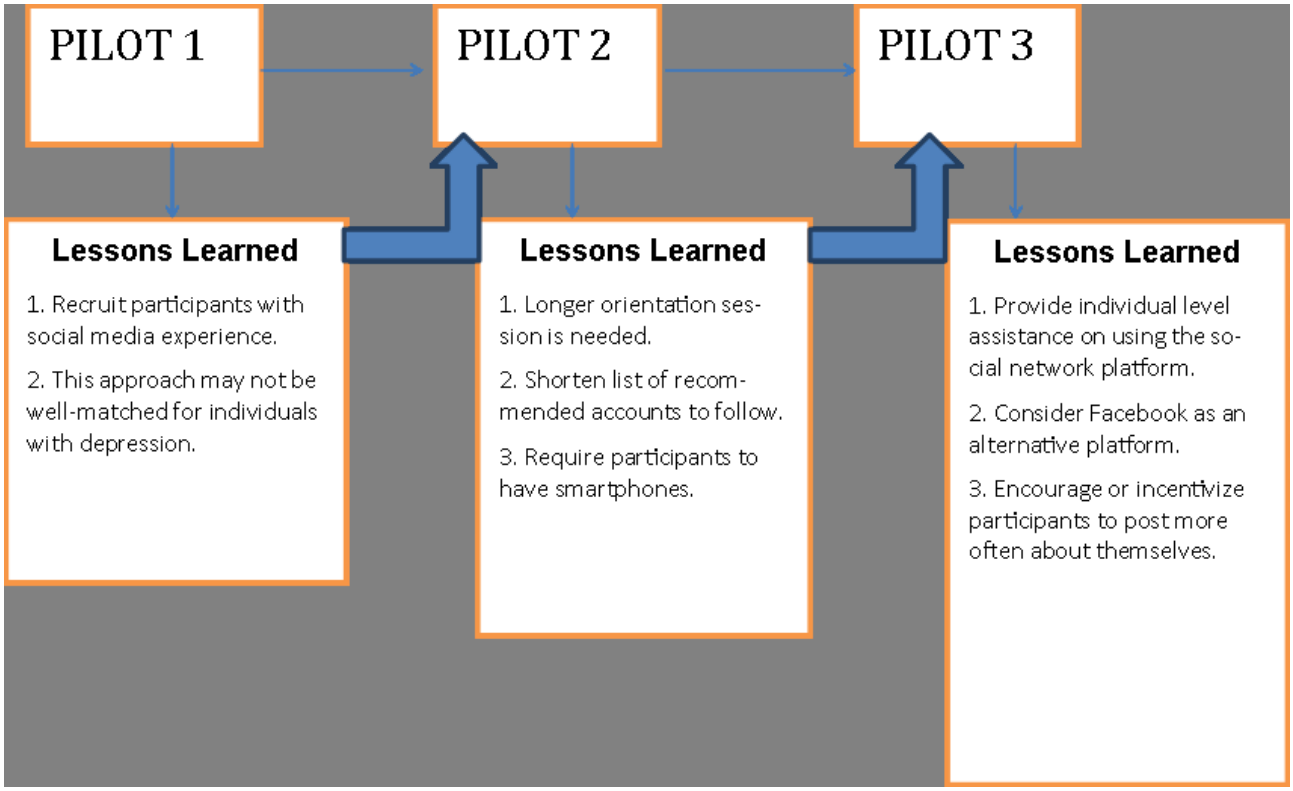
Lifestyle interventions (ie, behavioral weight-loss interventions) have had established efficacy for over a decade, but are still not widely disseminated largely due to high cost and patient and provider burden [1-3]. Online social networks provide an alternative mode for delivery of lifestyle counseling which may reduce patient visits, the main source of cost and burden of traditional modalities. Interactions in online social networks are frequent, brief, and asynchronous because users log into their online communities during downtime or when they simply feel a need for social connection. As such, social media has become embedded into many people's daily lives. Online social networks may then provide a means to embed health behavior-change programming into people's daily lives [4]. Another advantage of using social media to deliver lifestyle interventions is that it increases opportunities for patients to receive social support for their weight-loss efforts [5], which may be particularly important for socially isolated populations, such as those with depression [6].

Most studies have used an online social network as an adjunct to traditionally delivered weight-loss programs, either by conducting scheduled group chats online [7,8] or by providing a message board/forum for participants to submit questions and chat [9,10]. One study used an online social network as an adjunct to a podcast-delivered intervention but did not find that it improved outcomes relative to a podcast-only condition [11], while two ongoing studies are using an online social network as the main intervention delivery modality for weight-loss interventions [12,13]. Two recent systematic reviews of 12 and 20 studies, respectively, revealed that the impact of the online social network component of a weight-loss intervention has never been isolated [14,15]. Three studies showed that engagement in the online social network predicted greater weight loss, which suggests that the social network component may have a role in promoting better outcomes [16-18]. Although research on the role of online social networks in facilitating weight loss shows promise, further studies are required to determine the ideal way to utilize the social network (ie, as an adjunct versus the sole treatment modality), participants most likely to prefer this modality, and who will benefit the most.

Another gap in the literature is that few social network weight-loss studies have used mainstream social networking platforms like Facebook or Twitter [14,15]. Investigator-designed websites may lack the technological sophistication and usability of mainstream platforms, which have undergone many years of refinement by expert developers [14]. Leveraging widely used and freely available social networking platforms in health behavior interventions can increase the sustainability and dissemination potential of interventions, and eliminates the costs associated with developing new platforms that can quickly become obsolete.

This report describes an iterative series of three pilot studies in which we evaluated the feasibility and acceptability (eg, engagement and retention) of using Twitter, a mainstream commercial online social network, as an adjunct to a traditional group visit-delivered lifestyle intervention in depressed and nondepressed samples, and as the primary intervention modality in a nondepressed sample. This pilot series provided multiple opportunities to iteratively refine the intervention based on our experiences in each study. We present the methodology, results, and lessons learned for each study in the order they were performed to demonstrate how this line of work evolved (see Figure 1). The overarching goal of this work was to determine for whom online social networks might be most acceptable and how much of the intervention is feasible to deliver via the online social network. In each pilot, we created a private social community on Twitter as either an adjunctive component of an in-person lifestyle intervention or as the primary intervention modality. Focus groups were conducted after each pilot to get feedback on what participants liked and disliked about the social network aspect of the program. Survey items explored the acceptability of the social network and of various forms of engagement. Finally, to characterize the social influence of Twitter groups relative to other relationships, we compared participants' perceptions of social support and negativity regarding weight from their Twitter group against perceived social support and negativity in preexisting in-person relationships (ie, family and friends).

Figure 1. Flow of research.



Methods

Overview of Pilot Series

In Study 1, we enrolled adults with obesity and depression. Individuals with depression tend to have greater difficulty losing weight [19] and tend to feel more socially isolated [20]. A supportive social network could provide participants with social experiences devoid of weight stigma, which can increase depressive symptoms and inhibit weight loss [21,22]. The intervention involved 12 weekly visits and an adjunctive online social network component (via Twitter) that connected participants and counselors. In between group visits, counselors used Twitter to continue conversations about the topic of the week, share links to relevant content, provide support when participants reported progress or struggles, and interact with participants who missed visits. We hypothesized that individuals with depression would find the online social network an acceptable and desirable means of eliciting extra support in between group visits. Given the challenges reported by the sample in Study 1 and an aim to further increase generalizability, in Study 2 we enrolled adults who were not depressed and who had regular social media experience into the same intervention. Given the high level of acceptability of the online social network in this population, in Study 3 we enrolled participants with similar characteristics as those in Study 2 into a lifestyle intervention that was delivered almost exclusively via the online social network. Focus groups and surveys following each intervention evaluated acceptability and participants' likes and dislikes about the program. The University of Massachusetts (UMass) Medical School Human Subject Committee approved all study procedures.

Study 1 Methods

Recruitment and Screening

In August 2012, participants were recruited though the local community and the UMass Medical School. Study flyers were posted on billboards throughout the community and university. Online advertisements were posted on local advertisement sites and the university distribution list. The study advertisement was also distributed via recruitment-specific newsletters sent to the community. Individuals responding to ads were screened by phone for eligibility. Eligible and interested individuals were scheduled for a baseline visit lasting 90 minutes during which they provided written informed consent and their height and weight were taken. Participants also completed surveys including the following: demographics, Beck Depression Inventory II (BDI-II) [23], medical history, medications, and social support for weight loss (eg, Weight Management Support Inventory [WMSI]) [24].

Inclusion and Exclusion Criteria

Participants were required to have a BDI-II score of >13 (mild or greater depression), be 18 to 65 years of age, have a body mass index (BMI) of 30 to 45 kg/m^2, have written clearance from their primary care provider to participate, and have Internet access in their home. Participants were excluded if they had initiated an antidepressant medication within past 2 months, had plans to move during the study, were pregnant or lactating, had severe mental illness (eg, bipolar disorder or schizophrenia) or a BDI-II score >30, had bariatric surgery, had a condition that precludes lifestyle changes, were taking a medication affecting weight, had type 1 or type 2 diabetes, or were active Twitter users (ie, tweeted in the last 3 months). Recruiting both users and nonusers would have essentially created two

subgroups that, with a small sample, might not adequately capture the experience of either, and recruiting only active Twitter users would have drastically limited the sample since only 23% of adults use Twitter [25]. Additionally, Twitter is generally used as a public forum, thus active users would not be able to use their current accounts but instead would have to start a second account to participate in the study since participation required the use of privacy settings. To reduce this variability in the small sample, we recruited people not actively using Twitter.

Intervention

Participants were enrolled into a 12-week weight-loss intervention involving an orientation visit, 12 weekly group counseling visits, and access to a private online Twitter group. The orientation visit lasted 90 minutes and assisted participants with setting up a Twitter account and learning how to use the mobile app, MyFitnessPal [26], to track dietary intake and physical activity. Participants developed their Twitter profiles using avatars and pseudonyms and posted their first tweets. They were encouraged to (1) tweet questions to the group and group leaders, (2) participate in discussions (eg, "What is the biggest challenge you have with holiday eating?") and challenges (eg, "Post a pic you took while exercising outside!") put forth by group leaders, (3) report their exercise, and (4) share their victories and challenges. Participants were encouraged to follow a list of 50 other weight- and health-related Twitter accounts and were provided a list of such accounts that were vetted by the investigators to insure the legitimacy of information provided.

Group Visits

A shortened version of the Diabetes Prevention Program (DPP) Lifestyle Intervention [27] was delivered by a clinical psychologist and exercise physiologist over the course of 12 weekly, 90-minute group counseling visits. Participants were given a calorie goal based on their basal metabolic rate that was adjusted to create an approximately 1- to 2-pound weight loss per week. They were encouraged to increase their exercise gradually to 150 minutes per week of moderate-intensity physical activity. Each participant was weighed privately at the beginning of group visits.

Online Group

The group leaders posted discussion topics each day based on the topic of discussion in the group visits. They also posted relevant articles, healthy recipes, information about healthy community events (eg, 5K races), and launched challenges and quizzes. Group leaders also directly tweeted participants who had not engaged in the social network in 1 week by mentioning their names in tweets (eg, "@puppymama, how are you doing?") in order to draw the participants back into the conversation.

Measures

Weight

Weight was assessed using a digital scale (Scale-Tronix, Model 5002, White Plains, NY) with participants wearing light clothing.

Retention

A dropout was defined as an individual who exited the study before it ended, meaning they discontinued both group visits and posting tweets in the online group, and they failed to attend a follow-up assessment visit.

Online Social Network Engagement

Engagement was defined broadly as the average number of Twitter posts and/or replies made by a participant during the 12-week program.

Acceptability

Survey items were developed to evaluate how helpful the Twitter social network was as a source of support and information, how likely they would be to continue using Twitter after the study, and how much they liked various forms of engagement, including posting updates about themselves, asking questions, replying to others' questions, and reading others' posts. Responses were on a scale from 0 (not at all/not at all likely) to 10 (very much/very likely).

Weight-Loss Social Support

Participants were asked to rate weight-loss social support they experienced from three relationship categories: Twitter friends (ie, group members and anyone else they may have decided to follow), in-person friends, and family [28]. The following definition of in-person friends was given: "Any friends that you interact with in person, meaning you see them and spend time with them." Three of the four subscales of the Weight Management Support Inventory, a validated weight-loss social support scale, were administered [24]: the informational (seven items), emotional (six items), and appraisal (three items) subscales. While these three subscales have been shown to be valid in online weight-loss programs, the instrumental support subscale was not, so we did not include it [29]. We eliminated three items from the original appraisal scale because, as worded, they refer to physical observations of an individual (eg, "others tell me I look like I'm in better shape"), which is not possible in an online social network. Each subscale included both a frequency (ie, how often did this occur in in the past 4 weeks) and a helpfulness dimension (ie, how helpful was each). Frequency responses were on a 5-point Likert scale from *not at all* to *about every day*. The helpfulness responses were on a 5-point Likert scale from *not at all helpful* to *extremely helpful*.

We also asked questions regarding social support for weight loss and negative social influence using items found to be internally consistent in our previous work [28]. These questions used 5-point Likert scales with responses ranging from *very much disagree* to *very much agree*. Single-item questions were used to assess five different aspects of positive social support—comfort, helpfulness, support, information, and fun—in regard to people in each of the three relationship categories. As in previous research [28], items were summed to create a total score with higher scores reflecting more positive social influence. Two items were used to assess negative social influence, one that asked participants to rate how embarrassed they feel discussing weight-related issues with people in each relationship category, and another that asked participants to rate how judgmental they feel people in each relationship category

are when it comes to weight-related issues. These items were not combined into a composite because they did not have high internal consistency.

Focus Group

Analytic Plan

All outcomes were summarized with descriptive statistics. Two investigators (JO, SP) reviewed focus group responses and developed themes for each category of responses. Responses were then coded by two investigators (JO, RH) and discrepancies were discussed until consensus was reached. Quantitative analyses were conducted using SPSS version 22.0 (IBM Corp, Armonk, NY). Qualitative analyses were conducted using NVivo version 10.0 (QSR International, Doncaster, Australia).

A 1-Hour focus group led by an experienced focus group facilitator was convened after the 12-week follow-up visit to ask participants what they liked best and least about the online aspect of the program and their suggestions for improvement. A note taker was present and the focus group was recorded and later transcribed for the coders. Participants were compensated for attendance.

Study 1 Results

Overview

The sample (n=10) was 90% female (9/10), had a mean age of 46.2 years (SD 10.9), and was 80% (8/10) non-Hispanic white with a mean BMI of 35.86 kg/m^2 (SD 4.62) and a mean BDI-II score of 16.82 (SD 11.90) (see Table 1). Most participants (8/10, 80%) were employed full time. A total of 30% (3/10) of participants had no social media accounts, 40% (4/10) had one social media account, and 30% (3/10) had more than one social media account. Among those who used social media, 70% (7/10) reported logging into their accounts at least once per day.

Measures

Retention, Attendance, and Engagement

One participant dropped out, resulting in a 90% (9/10) retention rate. On average, participants attended 7.8 of the 10 group visits (SD 2.2; range 3 to 10). Participants posted an average of 110.7 tweets (SD 112.4; median 66.6, interquartile range [IQR] 167) over the course of 12 weeks.

Weight Loss

Participants lost a mean of 2.3 pounds (SD 7.7; range -19.2 to 8.2) or 1.2% (SD 3.6) of their baseline weight. A total of 2 out of 10 (20%) participants lost clinically significant weight (ie, ≥5% of baseline weight), 40% (4/10) were at the same weight as they were at baseline, and 20% (2/10) gained 1 pound or more (see Table 2).

Acceptability

Of the 9 participants that completed the acceptability survey, two-thirds (6/9, 67%) reported that they found the Twitter group to be at least a somewhat useful source of support and information and that they are at least somewhat likely to continue to use Twitter after the study has ended (see Table 3). Just over half (5/9, 56%) reported to at least somewhat like posting a status update about themselves or posing a question to the group. Two-thirds (6/9, 67%) reported to at least somewhat like replying to other people's questions and just over half (5/9, 56%) at least somewhat liked seeing other people's posts.

Weight-Loss Social Support

At follow-up, participants' scores on the WMSI emotional subscales for frequency and helpfulness were not significantly different by relationship category (*P*=.12 to .90), however informational support subscales for frequency and helpfulness were significantly higher for the Twitter group relative to family (*P*=.01 and .02, respectively) and friends (*P*=.02 and .01, respectively). Appraisal support scales for frequency and helpfulness were not significantly different by relationship category (*P*=.05 to .73). Participant ratings of weight-related positive social influence (*P*=.10) and negative social influence, including embarrassment (*P*=.08), and how judgmental the people are (*P*=.06) did not differ by relationship category (ie, Twitter group, family, and friends) (see Table 4).

Focus Group

A total of 7 out of 10 participants (70%) attended the focus group. Several themes emerged from a total of 16 responses about what participants liked most about the online aspect of the program. Major themes were each endorsed by 3 of the 7 (43%) participants and included the following: being in a group of people with shared interests and goals (eg, "I liked that we all had a common goal."), enjoying interacting with others (eg, "I like the social interaction and openness of the group."), and social support for weight loss (eg, "Support from the group was uplifting."). Minor themes, each endorsed by 2 out of 7 participants (29%), included the responsiveness of others when they posted (eg, "I liked the fast and immediate responses I got."), anonymity (eg, "No one knows who you are."), and the information they received (eg, "Lots of tips, good information, and recipes."). Finally, 1 participant out of 7 (14%) said they did not like anything about the online piece (eg, "I don't tweet.").

Several themes emerged from a total of 11 responses about what participants liked least about the online aspect of the program. The major theme, endorsed by 3 out of 7 participants (43%), was that there was not anything they disliked about the program. Three minor themes emerged, each endorsed by 2 of 7 participants (29%), and included difficulty understanding how to use Twitter, finding the feed to be overwhelming, and having limited access to Twitter due to work or lack of mobile phone. Finally, 1 participant of 7 (14%) mentioned that she did not feel comfortable posting about herself.

Lessons Learned From Pilot 1

Because participants in Study 1 lost very little weight on average and reported many barriers to using the online social network, we concluded that perhaps the extra effort of this aspect of the intervention was not adding value and even possibly detracting value. Previous research shows that individuals with depression have higher rates of treatment failure [19] and this study seemed to be no exception. Another factor affecting the perceived effort of using the online social network was the lack of social media

experience in this sample. Based on these results, for Study 2 we decided to keep the intervention the same but to enroll adults who did not have depression and who had a higher degree of social media experience as indicated by their being regular Facebook users. Facebook is the most highly used online social network [25], thus this inclusion criteria would not sufficiently hinder recruitment and addressed concerns we had regarding enrolling existing Twitter users who would then need to create a second private account.

Study 2 Methods

Recruitment and Screening

In April 2013, the same recruitment and screening procedures were used as in Study 1, except the BDI-II cutoff was not used.

Inclusion and Exclusion Criteria

Eligibility criteria included active Facebook and mobile phone users aged 18 to 65 years who had a BMI between 30 and 45 kg/m^2 and approval from their primary care physician to participate. Participants were excluded if they had plans on moving during the study period, were currently pregnant or lactating, had plans to have bariatric surgery during the study period, had medical conditions preventing dietary changes or an increase in physical activity, had type 1 or type 2 diabetes, and were taking medications associated with weight gain.

Measures

The measures used in Study 2 were the same as those used in Study 1.

Intervention

Group visits and the Twitter component were carried out in the same fashion as in Study 1.

Analytic Plan

The analytic plan was the same as that in Study 1.

Study 2 Results

Overview

Participants (n=11) were all female (100%) and largely Caucasian (9/11, 82%) with a mean age of 48.3 years (SD 12.4) and a mean BMI of 33.8 kg/m^2 (SD 3.7). Most (10/11, 91%) were employed full time. The mean BDI-II score was 5.4 (SD 3.9), which fell well within the minimal depression range (0 to 13). The majority of participants (10/11, 91%) had more than one social media account and 64% (7/11) of social media users reported at least daily log-ins (see Table 1).

Measures

Retention, Attendance, and Engagement

Two participants dropped out, resulting in an 82% (9/11) retention rate. On average, participants attended 8.09 of the 12 group visits (SD 3.60; range 1 to 12). Participants posted an average of 121.9 tweets (SD 127.0; median 73.0, IQR 191) over the course of 12 weeks.

Weight Loss

Participants lost a mean of 5.6 pounds (SD 6.3; range -15 to 0) or 3.0% of their baseline weight (SD 3.4) (see Table 2). A total of 36% (4/11) of participants lost clinically significant weight (ie, ≥5% of baseline weight), 27% (3/11) lost less than 5% of their baseline weight, and 36% (4/11) had no weight change. No participants gained weight.

Acceptability

Of the 9 participants who completed the acceptability survey, all (100%) found the Twitter group to be at least a somewhat useful source of support and the vast majority (8/9, 89%) found it to be at least a somewhat useful source of information (see Table 3). All participants said they would be at least somewhat likely to continue with Twitter following the intervention. The vast majority of participants at least somewhat liked posting a status update (7/9, 78%), posing a question (8/9, 89%), replying to others' questions (9/9, 100%), and reading other people's posts (9/9, 100%).

Weight-Related Social Support

At follow-up, emotional support subscales for frequency were significantly higher for the Twitter group relative to family (P=.01) and friends (P<.001). The emotional support subscales for helpfulness were not different between the Twitter group and family (P=.12), but were higher for the Twitter group than for in-person friends (P=.04). The informational support subscales for frequency and helpfulness were greater for the Twitter group than for both family (P<.001 and P=.01, respectively) and friends (P<.001 and P=.01, respectively). The appraisal support subscales for frequency and helpfulness did not differ by relationship category (P=.48 to .85). Positive social influence differed by relationship category (P=.005) (see Table 4). Participants rated their Twitter group as a greater source of positive social influence for weight loss compared to their family (P=.007) and in-person friends (P=.03). In terms of how embarrassed they felt discussing weight-related topics, no differences were found in participants' ratings across relationship types (P=.33). Differences emerged in ratings of how judgmental they rated each relationship category (P=.03), such that participants rated their family as more judgmental than their Twitter group (P=.003).

Focus Group

The focus group was attended by 9 of 11 participants (82%). Several themes emerged from a total of 22 responses about what participants liked most about the online aspect of the program. Five major themes emerged. The most frequently endorsed theme, endorsed by 7 of the 9 participants (78%), was social support received from the group (eg, "For me, I need positive feedback. People would say positive, nonjudgmental things."). The next major theme, endorsed by 5 of 9 participants (56%), was feeling nudged or inspired by others' posts (eg, "You see someone post they walked 3 miles and think, hey she's making me look bad! I can get off my butt and do that too!"). Liking that everyone was on the same path (eg, "It's nice to have people that I can talk to even though I don't spend time with them I feel like I know them, we all have the same goal.") was a major theme endorsed by 4 of 9 (44%) participants. Information (eg,

"I liked getting information and recipes right in front of me.") and instant feedback (eg, "Every time I posted a walk I would get a favorite or reply the same day and that felt good.") were major themes, each endorsed by 3 of the 9 (33%) participants.

Two major themes emerged from a total of 15 responses about what participants liked least about the online aspect of the program. The most frequently occurring theme, endorsed by 8 of the 9 participants (89%), was the Twitter feed being overwhelming with too many tweets from too many people they were following. The other major theme was the usability of Twitter (eg, "The app had different functions than on the computer."), endorsed by 4 of the 9 (44%) participants. A total of 3 of the 9 participants (33%) made unique comments that did not fit into the major themes. Out of the 9 participants, 1 (11%) mentioned feeling uncomfortable when strangers would send follow requests (eg, "I had this experience with some guy that started following me and I have no idea how he found me or why he would care. That bothered me."). Out of the 9 participants, 1 (11%) said she did not know what to post (eg, "I didn't think I really had anything to say.") and 1 (11%) said there was not anything she disliked.

Lessons Learned From Study 2

Given the more promising weight losses and the higher level of acceptability in Study 2 relative to Study 1, we concluded that adults who have experience with social media and no major mental health barriers may be a promising group to attempt a more aggressive social media-delivered approach. Thus, for Study 3 we decided to deliver a far greater amount of intervention content through the online social network and to limit in-person group visits to one. We required participants to have mobile phones and experience using mobile apps to insure competence in accessing apps from mobile phones and to maximize accessibility to the group. We also extended the orientation by 30 minutes and spent more time on Twitter functions. Finally, we decided against giving participants a list of 50 accounts to follow since they seemed to find the feed too overwhelming, but rather suggested just 10 and said following the accounts was optional.

Study 3 Methods

Recruitment

In October 2013, recruitment methods described for Study 1 were used.

Inclusion and Exclusion Criteria

Eligibility criteria included having a Facebook account, being aged 18 to 65 years, and having a BMI between 30 and 45 kg/m^2. Participants were excluded if they had used Twitter in past 3 months, had plans to move during the study, did not have a scale at home to weigh themselves, were currently pregnant or lactating, had plans to have bariatric surgery during the study period, had medical conditions preventing dietary changes or an increase in physical activity, had type 1 or type 2 diabetes, were taking exclusionary medications, were not a current mobile phone user, or had never used a mobile app.

Intervention

Orientation

Participants were enrolled into a 12-week weight-loss intervention involving an orientation visit, a midintervention visit, and a private online group formed using Twitter. The orientation visit lasted 2 hours and was similar to the one in Studies 1 and 2 except it involved additional practice at each step because our focus group results from Studies 1 and 2 suggested that participants wanted more instruction on how to use Twitter. Participants also received their calorie, physical activity, and weight-loss goals at the orientation session.

Online Group

Diabetes Prevention Program Lifestyle Intervention materials were converted into online articles and links were tweeted with a byline that described the content (eg, "8 tips for healthier eating out! Which will you try this week?"). Each day, group leaders posted a new topic with accompanying links and discussions. Counselors logged in twice a day to start discussions, answer questions, provide positive reinforcement, and share content. Participants were emailed each week to elicit their self-reported weights. As in Study 1, group leaders made daily posts of relevant articles, healthy recipes, information about healthy community events (eg, 5K races), and launched challenges and quizzes. Group leaders tweeted at specific participants who had not engaged in the social network in 1 week to draw the participant back into the conversation as in Studies 1 and 2. Participants were given a list of 10 suggested Twitter accounts to follow that included healthy recipe websites (eg, @cookinglight) or popular weight-loss bloggers, but were informed this was optional.

Midintervention Visit

At week 6, participants attended a group visit in which group leaders did "check-ins" and problem solving with participants regarding any challenges they were experiencing in their weight-loss journey or with participating in the Twitter group.

Measures

The measures used in Study 3 were the same as those used in Studies 1 and 2.

Sample

Participants (n=12) were largely female (11/12, 92%) and Caucasian (9/12, 75%) with a mean age of 45.8 years (SD 9.6) and a mean BMI of 34.1 kg/m^2 (SD 3.6). Most (10/12, 83%) were employed full time. Participants' mean BDI-II score was 5.6 (SD 6.2). Most participants reported having more than one social media account (10/12, 83%): 33% (4/12) reported daily log-ins and 42% (5/12) reported greater than daily log-ins.

Study 3 Results

Measures

Retention, Attendance, and Engagement

All participants provided weight data at the end of treatment. A total of 2 of the 12 (17%) participants failed to attend the follow-up visit but instead sent their weights via email. Only 6 participants out of 12 (50%) attended the week 6 visit.

Participants posted, on average, 130.3 tweets (SD 124.0; median 74.0, IQR 211) over 12 weeks.

Weight Loss

Participants lost a mean of 5.4 pounds (SD 6.4; range -14.2 to 3.9) or 3.0% of their baseline weight (SD 3.1). A total of 2 participants out of 12 (17%) provided weight by self-report. Of the 10 out of 12 participants (83%) who had weight measured objectively, mean weight loss was 5.04 pounds (SD 6.20; range -12.8 to 3.9) or 2.4% of their baseline weight (SD 2.9). A total of 42% (5/12) of participants lost clinically significant weight (ie, ≥5% of baseline weight), 33% (4/12) lost less than 5% of their baseline weight, none remained weight neutral, and 25% (3/12) gained weight.

Acceptability

Of the 10 participants who completed the acceptability survey, the vast majority of participants found the Twitter group to be at least a somewhat good source of support (9/10, 90%) and information (8/10, 80%) (see Table 4). The majority (7/10, 70%) said they would be at least somewhat likely to continue to use Twitter after the study. Most participants at least somewhat enjoyed all types of posts with 70% (7/10) reporting this for status updates, 80% (8/10) for posing a question, 80% (8/10) for replying to others' questions, and 90% (9/10) for reading others' posts.

Weight-Related Social Support

At follow-up, participants' ratings of frequency and helpfulness of emotional support were not different by relationship category (P=.30 to .53). Participants' ratings of frequency of informational support were greater for the Twitter group than for both family (P=.01) and friends (P=.02), while ratings of helpfulness of informational support were greater for the Twitter group relative to family (P=.01), but not friends (P=.08). The appraisal support subscales did not differ by relationship category (P=.39 to .79). Participants' ratings of positive social influence were significantly different across relationship categories (P<.001). Participants rated their Twitter group higher in positive social influence for weight loss than their family (P=.03), but not in-person friends (P=.21). Ratings of embarrassment (P=.11) and how judgmental (P=.40) each relationship category was were not significantly different.

Focus Group

The focus group was attended by 10 of 12 participants (83%). Five major themes emerged from a total of 35 responses about what participants liked most about the online aspect of the program. The most frequently occurring theme, endorsed by all 10 (100%) participants, was encouragement from the group (eg, "There was always something encouraging on there and nonjudgmental."). The second-most frequently occurring theme, endorsed by 9 of 10 (90%) participants, was feeling nudged or inspired by others' posts (eg, "Being inspired by other people's posts, like oh she went for a walk, alright I'll go too."). The next two major themes were both mentioned by 6 of 10 participants (60%) and included feeling not alone (eg, "It felt like a secret little society of people on the same path.") and receiving valuable and relevant information (eg, "I like how you get information like recipes without having to spend time searching the Web."). Finally, a major theme was instant feedback (eg, "When I went out for a walk and posted it, someone would always see it and favorite it. Instantly.").

One major theme and three minor themes emerged from a total of 20 responses on what participants disliked most about the online part of the program. The major theme, endorsed by all 10 (100%) participants, was not preferring to follow people other than fellow group members (eg, "Too many tweets in the stream made it hard to find the group tweets at times."). Minor themes, each endorsed by 2 of 10 (20%) participants, included difficulties navigating the functions of Twitter (eg, "I never used Twitter before, I found it difficult to navigate."), wanting clearer notification when someone has tweeted them ("I didn't know how to tell someone was tweeting me."), no dislikes at all, and privacy concerns (eg, "The thing with Twitter is that anyone can find you."). While the orientation reviewed navigation, notification functions, and privacy settings, participants were new to Twitter and the amount of information shared in the orientation may have been too much at once. For example, the participant with concerns about being found by anyone on Twitter must not have understood that by using the avatar and pseudonym, along with privacy settings where only users she approves can see her tweets, would all make it impossible for anyone to find her on Twitter. This was clarified when she expressed this concern. The 2 out of 10 (20%) participants who said there was nothing they disliked about Twitter were, not surprisingly, former Twitter users. Finally, 1 participant out of 10 (10%) did not like that the app and online Twitter interfaces differed, 1 (10%) participant did not like to be repeatedly mentioned in tweets when coaches checked in on her, and 1 (10%) mentioned not liking ads in the newsfeed.

Lessons Learned From Study 3

Although results were promising in Study 3, some participants still experienced barriers to using the Twitter interface. When using a novel platform, more extensive training in the platform is likely needed. Our orientation lasted 2 hours and was held in a group format which may not have met the needs of individuals who needed more intensive help. Individual orientation meetings and a prestudy trial period to make sure users achieve comfort with the modality might prevent usability issues. Another approach is to recruit users of the platform being used in the study. This would circumvent the learning curve of the social media platform. Previous studies have used both Twitter [30] and Facebook [31] in weight-loss interventions with success. Another lesson learned is that participants seemed more interested in hearing from each other than in following relevant sources or blogs. In our future work we will use our feed to push select resources and information to users rather than recommend they follow relevant feeds. This will also allow us to better moderate what content they receive, which may be important given how plentiful misinformation is on social media. Who a participant follows seems best determined by the participant, with some preferring very small networks, while others prefer growing their networks. We also learned that participants enjoy each other's posts, but at the same time many participants experienced anxiety about posting about themselves. In our future work we will examine whether incentivizing some participants to post regularly will increase engagement of others

via role modeling processes, but also enhance the group's experience since they enjoy hearing from each other. Future studies should explore novel ways to induce meaningful engagement that involves sharing of experiences.

Results

Table 1 shows the sample characteristics from the three iterative pilot studies. Table 2 shows the sample characteristics, weight loss, and social media engagement for each of the three interventions. Table 3 shows acceptability of the Twitter social network by participants. Table 4 shows the weight-related social support of participants by relationship category at 12 weeks.

Table 1. Sample characteristics of three iterative pilot studies.

Characteristics	Study 1 (n=10)	Study 2 (n=11)	Study 3 (n=12)
Age (years), mean (SD)	46.2 (10.9)	48.4 (12.3)	45.8 (9. 7)
Baseline BMI[a] (kg/m^2), mean (SD)	35.9 (4.6)	33.8 (3.7)	34.2 (3.6)
Female, n (%)	9 (90)	11 (100)	11 (92)
Caucasian, n (%)	8 (80)	9 (82)	9 (75)
Employed full time, n (%)	8 (80)	10 (91)	10 (83)
Ever used social media, n (%)	8 (80)	11 (100)	12 (100)
Has Facebook account, n (%)	6 (60)	11 (100)	12 (100)
Ever had Twitter account, n (%)	1 (10)	5 (45)	1 (8)
Have ever used online community for weight loss, n (%)	2 (20)	6 (55)	2 (17)

[a]Body mass index (BMI).

Table 2. Intervention and sample characteristics, weight loss, and engagement

Characteristics	Study 1 (n=10)	Study 2 (n=11)	Study 3 (n=12)
Intervention	12 group visits + Twitter	12 group visits + Twitter	1 group visit + Twitter
Length	12 weeks	12 weeks	12 weeks
Depression status	Depressed	Nondepressed	Nondepressed
Social media inclusion criteria	None required, not an active Twitter user	Daily Facebook use required, not an active Twitter user	Daily Facebook use required, not an active Twitter user
Technology inclusion criteria	Internet access at home	Internet access at home	Internet access at home, has a mobile phone
Weight change (lbs), mean (SD)	-2.3 (7.7)	-5.6 (6.3)	-5.4 (6.4)
Weight change (%), mean (SD)	1.2 (3.6)	3.0 (3.4)	3.0 (3.1)
≥5% weight loss, n (%)	2 (20)	4 (36)	5 (42)
Tweets, mean (SD)	110.8 (112.4)	121.9 (127.1)	130.3 (124.1)

Table 3. Acceptability of Twitter social network.

Questions from acceptability survey		Study 1 (n=9)	Study 2 (n=9)	Study 3 (n=10)
To what extent did you find Twitter to be useful as a source of support for your weight-loss effort?				
	Score, mean (SD)	3.0 (4.2)	8.0 (2.0)	6.8 (1.9)
	Rating a 5 or greater[a], n (%)	6 (67)	9 (100)	9 (90)
To what extent did you find Twitter to be useful as a source of information about weight loss?				
	Score, mean (SD)	5.5 (6.4)	8.2 (2.6)	7.2 (2.3)
	Rated a 5 or greater, n (%)	6 (67)	8 (89)	8 (80)
How likely is it that you will continue to use Twitter after the study has ended?				
	Score, mean (SD)	5.1 (4.1)	8.1 (2.4)	5.8 (3.2)
	Rated a 5 or greater, n (%)	6 (67)	9 (100)	7 (70)
How much did you like posting a status update about yourself?				
	Score, mean (SD)	4.9 (3.9)	6.0 (2.6)	6.6 (2.4)
	Rated a 5 or greater, n (%)	5 (56)	7 (78)	7 (70)
How much did you like posting a question?				
	Score, mean (SD)	4.7 (4.0)	6.9 (3.0)	6.3 (2.9)
	Rated a 5 or greater, n (%)	5 (56)	8 (89)	8 (80)
How much did you like replying to a question posed by someone else?				
	Score, mean (SD)	5.1 (4.2)	7.4 (1.9)	6.8 (3.1)
	Rated a 5 or greater, n (%)	6 (67)	9 (100)	9 (90)
How much did you like reading others' posts?				
	Score, mean (SD)	5.3 (4.0)	8.9 (1.8)	8.1 (2.4)
	Rated a 5 or greater, n (%)	5 (56)	9 (100)	9 (90)

[a]All response options were rated on a scale from 0 to 10 from *not at all (likely)* to *very much/likely*.

Table 4. Weight-related social support by relationship category at 12 weeks.

Social support		Study 1, mean (SD)	Study 2, mean (SD)	Study 3, mean (SD)
Emotional support: frequency				
	Twitter group	2.3 (0.9)	2.8 (0.5)[a]	2.6 (1.1)
	Family	2.3 (0.7)	2.0 (1.0)	2.3 (1.2)
	Friends	1.9 (0.7)	1.7 (0.6)	2.2 (1.0)
Emotional support: helpfulness				
	Twitter group	2.7 (1.2)	3.3 (1.0)[b]	3.1 (1.2)
	Family	2.5 (0.8)	2.4 (1.5)	2.6 (1.2)
	Friends	1.9 (0.7)	2.1 (1.2)	2.8 (1.3)
Informational support: frequency				
	Twitter group	2.7 (1.1)[a]	3.2 (0.7)[a]	3.1 (1.2)[a]
	Family	1.5 (0.5)	1.7 (0.8)	1.6 (0.5)
	Friends	1.8 (0.7)	1.7 (0.6)	1.8 (0.9)
Informational support: helpfulness				
	Twitter group	3.1 (1.5)[a]	3.6 (1.1)[a]	3.5 (1.3)[c]
	Family	1.5 (0.4)	2.1 (1.1)	1.9 (0.8)
	Friends	1.8 (0.7)	2.2 (1.2)	2.4 (1.2)
Appraisal support: frequency				
	Twitter group	1.8 (0.5)	2.2 (0.8)	2.3 (1.3)
	Family	1.8 (0.7)	2.1 (1.0)	2.1 (0.8)
	Friends	1.8 (0.8)	2.1 (1.1)	2.0 (0.8)
Appraisal support: helpfulness				
	Twitter group	1.9 (0.7)	2.8 (1.2)	2.8 (1.6)
	Family	2.1 (0.8)	2.5 (1.5)	2.8 (1.3)
	Friends	2.2 (1.4)	2.7 (1.7)	3.2 (1.2)
Positive composite score				
	Twitter group	19.2 (7.8)	21.6 (1.7)[a]	19.7 (3.4)[c]
	Family	14.8 (4.5)	14.4 (5.7)	15.7 (5.7)
	Friends	15.7 (5.3)	17.4 (4.9)	18.3 (4.6)
Embarrassment				
	Twitter group	1.5 (0.8)	2.6 (1.7)	1.9 (1.2)
	Family	3.0 (1.9)	3.9 (1.3)	3.3 (1.4)
	Friends	3.8 (1.6)	3.6 (1.7)	2.8 (1.3)
Judgmental				
	Twitter group	1.2 (0.4)	1.4 (0.7)[a]	1.8 (1.3)
	Family	3.2 (1.5)	3.3 (1.4)	2.7 (1.6)
	Friends	2.2 (1.6)	2.3 (1.3)	2.2 (1.3)

[a]$P<.05$ for family and friends.

[b]$P<.05$ for friends only.

[c]$P<.05$ for family only.

In summary, retention rates were high, ranging from 81 to 100% across the pilots. Participants tweeted, on average, 9.3 to 10.8 tweets per week. Although we did not track the distribution of the tweets across time, the rate of engagement was more than

one tweet per day over the 12 weeks. In terms of weight loss, participants with depression had the lowest rate (20%) of clinically significant weight loss (≥5% of baseline weight), while 36 to 42% of nondepressed participants lost clinically significant weight. The pilot that included only one intervention visit (Study 3) did not reveal a trend toward less weight loss compared to the studies that had weekly intervention visits (42% vs 20 to 36% losing clinically significant weight), which supports moving to the next step of a fully powered randomized trial to compare Twitter-delivered versus traditionally delivered intervention on weight loss. Across all three studies, participants rated their Twitter group as at least as good as, or a significantly greater source of, weight-related social support than their close ties (ie, family and friends). Focus groups for all three studies revealed similar major themes about what was liked most about the online social network, including social support, encouragement, and nudging. The major themes about what was liked the least about the online social network were also similar across studies, including feeling overwhelmed by too many feeds to follow and the usability of Twitter.

Discussion

Principal Findings

This series of studies showed that using a private Twitter group as an adjunct to an in-person behavioral weight-loss intervention is feasible and acceptable in a sample of adults with obesity who did not have depression and who were regular users of social media (though Twitter-naïve). Among adults with depression who had less social media experience, this approach did not appear to be very acceptable. Although providing participants access to each other and counselors via online social networks conceivably could provide greater opportunities for social support, participants appear to need a certain comfort level with this modality to actively engage. In each study, we provided participants with an orientation visit to teach them to use the online social network; however, previous experience using an online social network may be more instrumental to their engagement and success than study-provided training. In Study 1, participants with depression reported that they were not sure how to solicit social support in the online setting. Greater guidance may be needed to activate patients to solicit social support via this modality. For example, giving patients specific ideas on what to post, putting them in communities with highly active and engaging users that may serve as role models, and proactively drawing them into conversations to ignite engagement could be tested in future work.

Among nondepressed adults attempting to lose weight, using an online social network as either an adjunct or as the primary intervention modality appears to be both acceptable and feasible. Retention was 82% when Twitter was used adjunctively (Study 2) and 100% when Twitter was used nearly exclusively as the intervention modality (Study 3). Thus, we found no reason to believe that offloading content to an online social network negatively impacts retention. In fact, the online social network modality may be particularly conducive to retention given that the usual barriers to participation in visit-based programs (eg, weather, schedule constraints, and travel) are not barriers in an online social network-based program. The single intervention visit was not likely deemed important or necessary to participants as it was difficult to schedule and only attended by 50%, so did not likely add much value. Mean weight loss observed across the pilots that recruited nondepressed adults (5.5 lbs in 12 weeks or 0.5 lbs per week) was fairly comparable to that achieved in the Diabetes Prevention Program Lifestyle Intervention (ie, mean 14.33 lbs in 24 weeks or 0.58 lbs per week), especially considering the latter had a far higher percentage of males than the samples of our pilot studies (32% vs 4.3%), and men achieved greater weight loss than women [32]. A randomized controlled noninferiority trial evaluating whether a Twitter-delivered behavioral weight-loss intervention is not appreciably worse than the traditionally delivered version is a next needed step to explore the efficacy of this approach when conducted over a full year, the usual length of behavioral weight-loss interventions. Whether participants will continue to engage in a longer-term lifestyle intervention administered entirely via an online social network is unknown.

Future research should explore whether adding a social network to a traditional behavioral weight-loss intervention improves weight-loss outcomes relative to a traditional version that does not include a social network. It remains unclear if the social network adds value or if it just adds more burden to an already burdensome intervention. Studies could explore the influence of the social network on social support and on attendance to group visits. One possible unexpected outcome could be reduced attendance at group visits to the extent that participants feel they can get enough information and support from the social network alone. Our study could not address this question directly, but we did observe poor attendance at the single study visit offered in the third pilot.

Findings extend previous research that showed that people using Twitter to talk about their weight loss rated their Twitter connections as more supportive and less judgmental than family and friends [28]. In Studies 2 and 3, we found that that the weight-related support participants experienced from their Twitter group exceeded that which they experienced from family and/or friends. The accessibility of an online group may increase opportunities for individuals to receive social support and the shared goals may strengthen the support received, even if these are loose social ties. Future studies using online social networks should examine ways to further enhance users' experience of social support, as this would be particularly instrumental for individuals who receive insufficient support for their weight-loss efforts elsewhere in their lives [5,28].

Behavioral interventions delivered via online social networks require the translation of intervention materials into a format that resembles communication habits of the social network. We created a content library that included online articles, brief headlines to be used in tweets to entice participants to click on and read the articles, exercise videos on YouTube, links to recipes from reputable sources, and other online resources. We developed orientation materials to advise participants on what to expect and how to interact when using this modality, and trained counselors in “microcounseling,” a way to deliver counseling in a brief, asynchronous manner each day as opposed

to in discrete chunks as is characterized by 60- to 90-minute visits that occur once a week.

Emerging research is shedding light on the type of content of posts most likely to be viewed, clicked, and read in weight-loss social networks. One study showed that polls, suggestions, and posts querying participants' weight-loss progress received the highest levels of participant engagement in a Facebook weight-loss group [8], and another found that polls and photos received the highest levels of engagement in a Facebook weight-loss group [33]. Research is needed to explore the timing of posts most likely to lead to clicks on links and the length of articles and videos that are most likely be read and viewed. To be successful in changing behavior, the online social network modality will require great attention to these issues. In our iterative series of pilot studies, where Study 1 informed Study 2, which then informed Study 3, we gathered qualitative and experiential data, and we continue to iterate these features in subsequent studies to identify best practices using online social networks as a behavioral intervention modality.

Future work should explore the best means for leveraging online social networks for weight loss in patients with depression. Barriers to engagement should be identified. Although none of our participants discussed worsening depressive symptoms or suicidality in the online social network, procedures for handling such crises should be established in advance. Participants were advised to call us if they had a crisis to report as opposed to posting it online or in a private message. Counselors logged in twice a day, 7 days a week, to closely monitor participants' posts. We recommend that counselors have a frequent and regular presence in social network-delivered interventions, not only to monitor posts but to stimulate engagement.

Limitations

This study has limitations. Our samples were predominantly female and Caucasian. A review of male representation in weight-loss trials revealed that, on average, only 27% of samples across trials are male [34]. Our study showed even lower rates of males with 2 males across all 33 participants (6%). It is notable that our male participants were among the lowest engagers, with one tweeting twice (he had no previous experience with social networking, no mobile phone, and reported only using his computer for email) and the other only seven times (he reported that he was hoping for more competition-style programming and would have preferred Facebook). Research is needed to determine how best to design social media-delivered programs for men, as this type of program may not be the best fit.

Because of challenges in utilizing the Twitter modality, we only included participants in Studies 2 and 3 who were current users of any other online social network, which limits generalizability. However, research from the Pew Internet & American Life Project found that 74% of online Americans have at least one social media account [23]. In all studies, most individuals had no experience with Twitter, which may have affected their comfort level in engaging with and using this platform. Focus group data revealed that problems understanding the Twitter interface was a barrier for some participants. Future studies might find better results by recruiting users of the same online social network used to deliver the intervention. By doing so, participants would not have to initiate a new social network habit (ie, checking another feed), time would not have to be allocated to training on the social network, and intervention content would appear in their usual feed. A potential challenge of using commercial online social networks is the lack of control over the user interface and unexpected changes in settings and features. We experienced no problematic changes in settings and features in our pilot series, but these could arise in future interventions and treatment providers must remain vigilant of these changes so they do not impact participant confidentiality. The challenges of using investigator-developed platforms include lack of experienced users, loss of opportunity for content to appear in participants' regular social media feeds, and development that will require a great deal of time and money.

Conclusions

Using commercial online social network platforms like Twitter to deliver behavioral weight-loss counseling may be a less expensive and more convenient alternative to traditional modalities that require numerous clinic visits. Findings have broad implications for behavioral interventions as this modality could be used for a variety of interventions provided that materials are appropriately translated for delivery in an online social network modality. Future research is needed to refine the populations for whom this modality is ideal, to test the efficacy of interventions delivered entirely via online social networks, and to design and deliver content in the most engaging and effective ways using this modality.

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Conflicts of Interest

None declared.

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Abbreviations

BDI-II: Beck Depression Inventory II
BMI: body mass index
DPP: Diabetes Prevention Program
IQR: interquartile range
NIH: National Institutes of Health
UMass: University of Massachusetts
WMSI: Weight Management Support Inventory

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Original Paper

A Novel Health Information Technology Communication System to Increase Caregiver Activation in the Context of Hospital-Based Pediatric Hematopoietic Cell Transplantation: A Pilot Study

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Abstract

Background: Pediatric hematopoietic cell transplantation (HCT), commonly referred to as blood and marrow transplantation (BMT), is an intense treatment modality that requires the involvement of engaged caregivers during the patient’s (child’s) prolonged hospitalization. The ubiquity of electronic health records (EHRs) and a trend toward patient-centered care could allow a novel health information technology (IT) system to increase parental engagement. The paucity of research on acute care, hospital-based (inpatient) health IT applications for patients or caregivers provides an opportunity for testing the feasibility of such applications. The pediatric BMT population represents an ideal patient group to conduct an evaluation due to the lengthy inpatient stays and a heightened need for patient activation.

Objective: The primary objective of this study is to assess the feasibility of implementing the BMT Roadmap in caregivers as an intervention during their child’s inpatient hospitalization. The BMT Roadmap is an inpatient portal prototype optimized for tablet with a user-centered design. It integrates patient-specific laboratory and medication data from the EHR in real-time and provides support in terms of discharge goals, home care education, and other components. Feasibility will be proven if (1) the BMT Roadmap functions and can be managed by the study team without unexpected effort, (2) the system is accessed by users at a defined minimum threshold, and (3) the qualitative and quantitative research conducted provides quality data that address the perceived usefulness of the BMT Roadmap and could inform a study in a larger sample size.

Methods: This will be a single-arm, nonrandomized feasibility study. We aim to enroll 10 adult caregivers (age ≥ 18 years) of pediatric patients (aged 0-25 years) undergoing autologous (self-donor) or allogeneic (alternative donor) BMT. Assenting minors (aged 10-18) will also be invited to participate. Recruitment of study participants will take place in the outpatient pediatric BMT clinic. After signing an informed consent, the research study team will provide participants with the BMT Roadmap, available on an Apple iPad, which will used throughout the inpatient hospitalization. To measure the study outcomes, approximately 6-8 semistructured qualitative interviews will be conducted periodically from pre-BMT to 100 days post-BMT and an additional 15-20 semistructured interviews will be conducted among BMT health care providers to assess perceived usefulness and usability of the system, as well as any associated workflow impacts. Quantitative survey instruments will only be administered to adult participants (age ≥ 18 years).

Results: Recruitment will begin in September 2015, and preliminary findings are expected in 2016.

Conclusions: This protocol offers a framework for the design and analysis of a personalized health IT system that has the potential to increase patient and caregiver engagement in acute care, hospital-based contexts.

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KEYWORDS

health IT; caregiver; activation; engagement; pediatric; hematopoietic cell transplantation

Introduction

A growing trend in patient-centered care and health information technology (IT) systems for self-management has led to innovation and better outcomes [1,2]. However, the inpatient setting has seen little advancement in this realm of patient or caregiver engagement. An opportunity exists to capitalize on the ubiquity of electronic health records (EHRs) to provide patients and caregivers real-time access to their own data in the hospital setting [3]. Hematopoietic cell transplantation (HCT), commonly referred to as blood and marrow transplantation (BMT), is an intense treatment modality that requires patients and caregivers to be engaged throughout the prolonged hospitalization, which can last up to 6 weeks. Due to the high-risk and long-term nature, pediatric BMT represents an ideal population for testing the feasibility of novel health IT systems.

Health Information Technology-Mediated Systems

Health IT systems used during hospitalization, particularly in high-risk diseases such as BMT, could improve clinician-participant engagement [2,4]. Such systems offer the potential to overcome constraints in health care delivery limited by provider time, complicated health information, and financial pressures. Now that the vast majority of acute care hospitals in the United States have an EHR [3], a tailored or personalized health IT system is an accessible option to engage caregivers with their child's health information [2]. The use of systems that integrate with EHR data would allow for caregiver empowerment in the BMT and other clinical environments. However, there remains a paucity of research on the use of health IT tools in hospital-based contexts. Utilizing qualitative research on the needs of the BMT caregiver population, this study supports a feasibility evaluation of a health IT system, the BMT Roadmap.

Blood and Marrow Transplantation

BMT is a potentially curative therapy for many malignant and nonmalignant hematologic conditions [5]. Despite advances over the past decade, which have led to improved outcomes, BMT is an intense treatment modality often of last resort for many conditions. A prolonged hospitalization is typically required, keeping patients and their caregivers at the hospital for up to a month or longer [6]. Patients often have a compromised immune system for months following discharge from the hospital, requiring vigilant disease management by families because of its associated high risk for transplant-related mortality [7].

Patient and Caregiver (Participant) Engagement

Effective communication between clinician-participants is essential in the delivery of health care and is known to impact clinical outcomes [8-10]. Research has shown that participants who are more activated are more likely to engage in disease-specific self-management behaviors and communicate more effectively with providers [11,12]. Moreover, an environment that supports the role of self-concept is more likely to have effective self-managers [13,14].

For critically ill children, such as in the BMT population, caregiver participation is essential [12]. In fact, BMT treatment mandates an identified caregiver [7]. Caregivers, the majority of whom are the mothers of the patients, often take a highly active role in supporting their children throughout the course of hospitalization and beyond [15]. The severity of the illness and high risk of the BMT treatment creates a highly complex self-management environment. While helping their children navigate treatment, caregivers often face major challenges in their social, financial, and emotional lives [16]. This highlights the importance of effective strategies to increase patient-caregiver activation through caregiver empowerment [11]. Empowerment can be increased by reducing the asymmetry of health information between health care providers and caregivers [8,17], which follows a trend toward patient-centered care [18,19].

Methods

This is a single-arm, nonrandomized study that will take place during the period of hospitalization for either allogenic or autologous BMT. Participants will be provided with the BMT Roadmap system at the time of admission and will be allowed to use it during their length of stay.

Study Design, Development, and Prototype Testing

The design, refinement, and development of the BMT Roadmap were based on user-centered design techniques that incorporated feedback from patients, caregivers, and health care providers [20-23]. Previous ethnographic research incorporated the exploratory evaluation of original concept wireframes to optimize functionality and design (Figure 1) [21,23]. The BMT Roadmap addresses 3 areas in which increased access to information may reduce stress experienced by parent caregivers: navigating the health system and medical communications, managing the day-to-day challenges of routine care, and transitioning to long-term outpatient management. The BMT Roadmap addresses these with 5 content modules personalized to the patients: (1) laboratory studies, (2) inpatient medications, (3) enrolled clinical trials, (4) list of the health care team, and (5) criteria for discharge. The criteria for discharge are presented via both the general progression of inpatient recovery (the "phases" of the BMT Roadmap) and the direct checklist (Table 1). The overall user interface metaphor of a "road map" was selected to represent the experience of BMT patients and caregivers, which encompasses multiple, distinguishable periods. It also references the intended purpose of the tool, to assist caregivers in navigating their child's BMT journey (Figure 2).

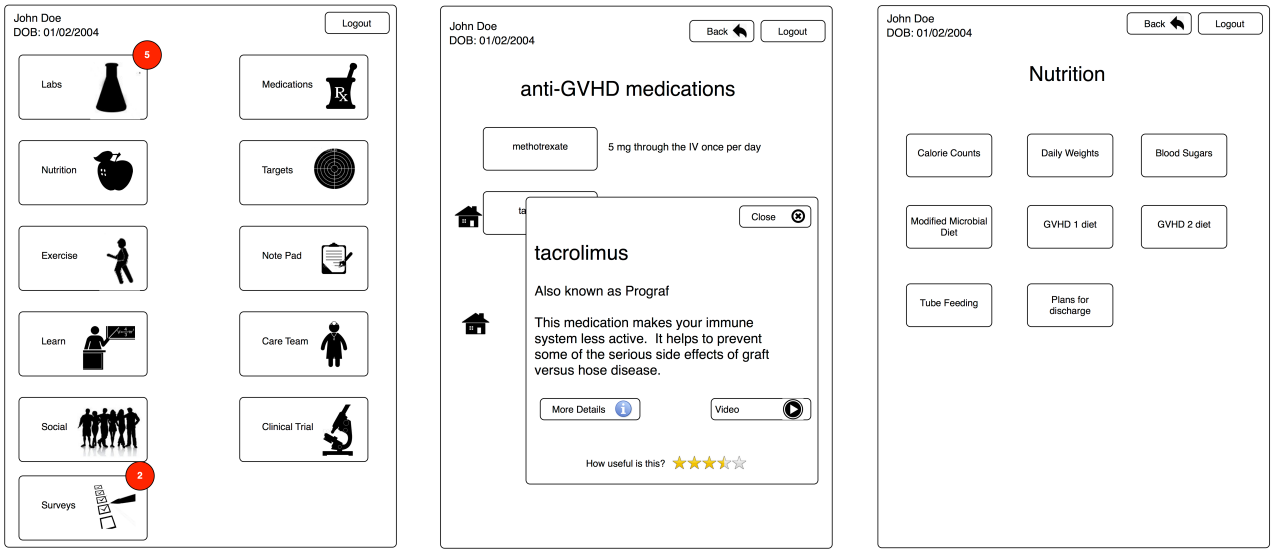
The BMT Roadmap is a Web application that integrates patient-specific data from our vendor EHR, Epic via Web services (Epic Systems Corporation, Verona, WI, USA). Following the wireframes informed through ethnographic research, design and programming work was done by the Center for Health Communications Research (CHCR) at the University of Michigan. During the development, certain functionalities were selected due to prioritized information areas, need for additional research, and feasibility of creation within available resources. Specifically, the BMT Roadmap tool that will be studied under this protocol does not include customizable goals, nutrition, or activities tracking that were explored in early stage, low-fidelity prototypes. The team felt that the selected functionality for this version of the BMT Roadmap represented the most important discovered information areas for the period of hospitalization and allowed for development within a reasonable use of resources. Future research and development will address the additional desires and areas of information management relevant to our patient-caregiver population. The application was optimized to be displayed on and used with an Apple iPad Air. The Apple iPad was chosen based on its familiar gesture-based interface, with which all of the BMT Roadmap operates, and its ergonomics. Additionally, our hospital's medical IT department could provide support for securely connecting an Apple iPad to our internal hospital wireless network. In addition to the CHCR team, development was collaboratively directed by the research team that encompasses several disciplines including expertise in BMT, computer-supported cooperative work, human-computer interaction, health informatics, survey methodology, and biostatistics.

Two "design workshops" (usability testing) were conducted (unpublished data, March and April 2015) to assess heuristic usability and perceived usefulness of the BMT Roadmap in patients and caregivers. A total of 10 families participated, including 10 caregivers and 7 patients. All families had previously been through the initial hospitalization associated with the BMT procedure and were currently receiving outpatient care. In Workshop 1, participants were given low-fidelity paper prototypes [23]. In Workshop 2, participants evaluated high-fidelity prototypes (wireframes on iPad devices). Participants were directly asked to respond to the user interface metaphor and overall tone of the BMT Roadmap. Overall, responses were positive and the team felt comfortable utilizing the metaphor in the final BMT Roadmap, with the intention to further consider user opinions under this protocol. The 5 modules (eg, labs, medications, clinical trials, health care providers, and discharge criteria) were tested in these families and additional input was sought related to information and caregiving themes. Observation notes and transcriptions from audio recordings were reviewed and discussed by the research study team (MM, DAH, EK, MA, LA, and SWC). Minor changes in the design, language, and functionality were made to reflect expressed concerns in usability. Major changes were documented as potential future functionality or modules not represented in this pilot BMT Roadmap system.

Table 1. Summary of the module features and addressed information areas.

BMT Roadmap module	Features of module	Addressed information areas		
		Navigating the health system and medical communications	Managing the day-to-day challenges of routine care	Transitioning to long-term outpatient management
Laboratory studies	Synchronously updated labs include complete blood count for entire hospitalization and a 3-day trend for electrolytes and liver panel, reviewed during rounds	X	X	X
Inpatient medications	Details of medications and their purpose, potential side effects, administration modes, and alternative names	X	X	X
Enrolled clinical trials	Includes plain-language summary and scan of the complete signed consent form, including length of participation and description of relevant tasks or specimens	X		X
List of health care team	Ordered alphabetically or by role, includes photo, name, and role	X	X	
Criteria for discharge (home screen)	Description of the “phases” of BMT that lead toward discharge (what to expect and when)	X	X	X
	Educational videos regarding practices to be done after discharge (safe maintenance of a central line catheter)			X
	Checklist of discharge criteria with bidirectional, 4-point progression slider; intended to set both expectations and task-oriented goals	X	X	X
	Glossary of common terms, available in a list and hyperlinked throughout content	X	X	X
	My Characteristics PDF—a patient-specific summary document frequently referenced by the medical team	X	X	

Figure 1. Original concept wireframes.



Description of Content Features

Clinical Phase Descriptions

Starting with chemotherapeutic conditioning before transplant, the BMT Roadmap lays out a progression that patients and caregivers can expect to go through during their HCT experience for families. This is done using graphics of a road map with buildings along the route as a visual metaphor for the major clinical events to be encountered during the treatment course. Each building has a description of potential symptoms or side effects and recovery techniques. By setting expectations in current and future phases, the aim is for families to have more advanced knowledge of the expected course of treatment and the progress one must make before discharge (Figure 2).

Patient Characteristics

Each patient will have a personalized "My BMT Characteristics" section that is developed by the health care team. This form will provide information unique to the patient undergoing BMT, such as type of conditioning chemotherapy regimen, dates of the procedure, infectious disease markers, blood type, etc.

Discharge Criteria

Discharge progress is represented alongside phases in the BMT Roadmap, describing general health and emotional and progress expectations. Discharge criteria are presented in the form of a to-do list. The caregivers will have the ability to note their progress for each element to mark when they are closer to completing the criteria using a bidirectional, 4-point sliding scale to reflect the fluidity of medical progress. Items also include educational tools, including self-care instructional videos for posthospitalization (Figure 2).

Medication List

All prescribed inpatient medications are displayed, grouped by purpose (eg, pain control, antibiotic, antigraft-vs-host disease), including the dose, method of administration, and side effects.

Lab Results

Lab results are updated from the EHR in real-time and offer basic visualizations of trends, as well as the most recent result in numeric form. The primary results of importance to these patients are provided including blood counts, and electrolyte and liver panels.

Medical Team

Because families have difficulty identifying and remembering the numerous people providing inpatient care throughout the day, we provide names and photographs of the care team, including roles (eg, nurse practitioner, attending physician, fellow).

Clinical Trials

Families also often forget what clinical trials they are enrolled in. Therefore, individualized clinical trial enrollment information can be provided for each patient, including a descriptive overview of the study, study details, and a scan of the consent form that was signed.

Figure 2. BMT Roadmap home screen, user interface metaphor and discharge criteria.

Glossary

The glossary includes commonly used terms and their definition. The glossary can be accessed on the home screen or by clicking on hyperlinked terms throughout the application.

Security Measures

Health information data displayed by the system will include patient laboratory results, medications, and a copy of signed informed consent documents. While this protected health information will be displayed on the iPad, the data are not stored

anywhere within the system. Rather, the latest results will be retrieved in real time using Web services to the EHR and displayed to the user. Users are authenticated using a username and password. Passwords are stored in the system using a secure, 1-way salted hashing function. Access to the underlying database is restricted to the host server only. Detailed audit trails of use are maintained. Servers will be hosted by the health system's medical center information technology (MCIT) department behind the institution's firewall, within the highly secure medical center data center. The iPads themselves are specially configured by MCIT to automatically connect to the hospital's secure, encrypted wireless network and to limit access to certain applications and URLs within the Web browser; users will not be able to install their own apps. This device will not work if removed from the hospital wireless network and can be deactivated remotely. Importantly, the security provisions for this application were thoroughly reviewed and vetted by the medical school's information services department using a formal risk assessment template approved by the health system's compliance office.

Patient and caregiver accounts will be created by the study team users through a secure management console accessible only by staff users. Patient and caregiver accounts are generated for users who have consented to be a part of the study, allowing for user-created password and a security question. The application times out after 10 minutes, requiring re-entry of the password. After 4 unsuccessful password attempts, account reactivation requires an administrator override. All functions require an authenticated user. Protected health information will be identified with access only provided to authorized personnel. The BMT Roadmap information system does not have any trust relationships with external environments (eg, interconnection agreements with third parties).

Participant Eligibility, Recruitment, and Accrual

Participants will be limited to those hospitalized for a first experience with BMT. Eligibility includes cancer and noncancer, autologous or allogeneic transplant cases. Any consenting adult caregiver (age ≥18 years) or minor assenting patient (aged 10-18 years with caregiver consent) is able to participate in the study. Based on the annual BMT census at the University of Michigan, 10 caregiver participants or patient-caregiver pairs (in the case of an assenting minor) will be recruited at the rate of 2-3 participants or pairs a month. All candidates for BMT are discussed at a weekly new patient evaluation meeting, which will help the study team identify potential study participants.

Health care providers who work in the pediatric BMT unit will also participate in research observation and interviews related to their experience as patients or caregivers use the BMT Roadmap.

Objectives

We hypothesize that the BMT Roadmap could provide a platform to promote caregiver (parent) activation and enhance health communication in a hospital-based context. Feasibility of the BMT Roadmap will be evaluated by considering outcomes of the following objectives:

1. Test the rate BMT Roadmap use in this population, with an expected threshold of 20% caregiver or patient enrollment into the study, with use throughout their hospitalization, based on at least a single login per day threshold.
2. Evaluate the impact of the BMT Roadmap system on caregiver activation, satisfaction, and burden through use of a survey instrument (eg, parent-patient activation model). To account for potential confounding, anxiety, mood, stress, miscarried helping, and experiential avoidance will also be measured through validated survey measurements instruments (Table 2).
3. Evaluate the usefulness and usability of the BMT Roadmap (in participants and health care providers) through a validated survey instrument (Table 2).
4. Assess the attitudes and perceptions of the participants and health care professionals on the BMT Roadmap through qualitative interviews.
5. Identify the presence and quality of care process redesign associated with the BMT Roadmap.

Table 2. Survey instrument summary.

Measures	Instrument	Description	Population	Time (min)	Baseline	Discharges	Day 100
Activation	Parent-PAM [12]	Modified version of PAM, which was developed to study parental activation using physical, emotional, and role domains of general functioning	Caregivers	5-7	X	X	X
Satisfaction	Press Ganey [24]	Patient inpatient experience and overall satisfaction	Caregivers	5-7		X	X
Anxiety	State Trait Anxiety Inventory [25]	Commonly used and validated tool with good test-retest reliability	Caregivers	5	X	X	X
Mood	Profile of Mood States [26]	Assess transient distinct mood states with 6 factor-based subscales: tension/anxiety, depression/dejection, anger/hostility, fatigue/inertia, vigor/activity, and confusion/bewilderment	Caregivers	5-7	X	X	X
Burden	Caregiver Quality of Life—Cancer [27]	35-item rating scale measuring physical, emotional, family, and social functioning burden will also be used to evaluate caregiver burden	Caregivers	8-10	X	X	X
Stress	Impact of Event-Revised [28,29]	22-item scale measuring difficulty faced over the past 7 days; the score may be reflective of an acute stress disorder	Caregivers	5-7	X	X	X
Miscarried helping	Helping for Health Inventory [30]	15-item scale to assess how caregivers perceive their own communication style regarding the patient's chronic illness; assesses miscarried helping (ie, how the well-intentioned efforts of caregivers of children with chronic diseases may become barriers to successful treatment)	Caregivers	5	X	X	X
Experiential avoidance	Parental Acceptance and Action Questionnaire (PAAQ) [31]	15-item scale that assesses experiential avoidance within the caregiver role. The PAAQ includes 2 factors: "inaction," composed of 9-items; and "unwillingness," composed of the remaining 6-items	Caregivers	5	X	X	X
Usefulness	Usefulness [32]	Custom questionnaire, as there are no validated instruments for measuring satisfaction specific to the BMT Roadmap information system	Caregivers, health care professionals	2	X	X	X
Usability	Usability [32]	Custom questionnaire, as there are no validated instruments for measuring satisfaction specific to the BMT Roadmap information system	Caregivers, Health care professionals	2	X	X	X

Outcomes Measures

Qualitative and quantitative research methods will be used to evaluate the objectives. From the time of admission until 100-days post-transplant, which is considered an end point for early recovery from HCT, the research team will conduct semistructured, qualitative interviews and administer validated surveys (Table 2). All interviews will be audio recorded.

Approximately 6-8 semistructured interviews per participant will probe around broad caregiver information needs, the perceived usefulness of the tool, and any literacy or usability feedback regarding the tool or its presentation on an Apple iPad. Participants will be encouraged to "think aloud" and will be prompted to complete particular tasks or offer understanding of specific information based upon previous interviews. Survey instruments allow for evaluation of outcome measures, such as usability of the tool and caregiver activation level, as well as potentially confounding factors, such as stress or mood (Table 2).

Another 15-20 qualitative interviews will be conducted of BMT health care providers, including physicians, nurses, pharmacists, nutritionists, social workers, and psychologists. Health care professionals will complete a subset of the surveys. Both interviews and surveys will seek opinions on the usefulness or usability of the tool from a provider's perspective, as well as the presence and quality of care process redesign in response to patients' or caregivers' use of the tool.

Additional data collected include demographic and baseline information, comprehensive system usage logs (eg, login frequency, pages viewed on the BMT Roadmap information system, time stamps) and clinical outcomes (eg, length of stay, risk of day 30 and 100 readmission, infections, transplant-related mortality, and survival).

Analyses

Qualitative interviews will undergo systematic, transcript-based analyses. Observational field notes will complement the recorded audio. Each session will be transcribed and coded to identify trends, adding additional codes and adjusting interview structure based on initial results.

Analysis of the survey instrument data will include a reference population of Parental-PAM (P-PAM) scores available in the pediatric BMT caregiver population that measures activation/participation. Comparisons of proportions of the study population to the reference population will be taken into account. Descriptive statistics will be calculated for each P-PAM score and stratified into the appropriate level of activation. Results will be compared with a published sample [12]. Univariate analyses will be performed to assess associations between P-PAM and demographic, social, and environmental characteristics of the parent (eg, type of insurance, marital status, number of children in household), disease-related characteristics of the patients (eg, age, disease, disease status at BMT), satisfaction [24], usefulness and usability [32], caregiver burden [27], mood [26], anxiety [25], stress [28], miscarried helping [30], and experiential avoidance [31]. Pearson's correlation (and other suitable measures of association for categorical variables) will be used to determine the nature and significance of association between each variable and Parent-PAM scores. Despite limited degrees of freedom due to the sample size, the survey instrument data will be analyzed using methods appropriate to the scale of items.

An exploratory analysis of system usage logs and clinical outcomes will be conducted to find potential correlations worthy of further study or that would inform future design of a personalized information system.

Pilot Study, Sample Size, and Retention

A sample size of 10 adult caregivers allows for an exploratory and feasibility study on the use of the BMT Roadmap at our institution. Recognizing that people have varying levels of technology skills, retention will be determined by demonstrating maintenance of at least 60% of patients and caregivers who agree to using the BMT Roadmap throughout inpatient hospitalization (ie, they log in at least once daily). By conducting interviews and surveys during routine hospital and outpatient care, there are minimal anticipated missing data. Given the sample size and the single-arm nature of the study, we will not be powered to determine statistically significant results in many outcome measures. However, the data within an approximate 1-year study period are expected to inform a future clinical trial and improved design of the BMT Roadmap.

Results

Recruitment will begin in July 2015 and results are expected in 2016. The evidence will reveal the feasibility of utilizing an educational health IT tool in the BMT hospitalization process.

Discussion

The goal of this pilot study is to evaluate the feasibility of implementing a patient-centric, personalized health IT system in the hospital-based BMT setting. In this case, feasibility will be proven if (1) the BMT Roadmap functions and can be managed by the study team without unexpected effort, (2) the system is accessed by users at a defined minimum threshold, and (3) the qualitative and quantitative research conducted provides quality data that addresses the perceived usefulness of the BMT Roadmap and could inform a study in a larger sample size. We hypothesize that caregivers will choose to use the BMT Roadmap, report overall satisfaction with the system, and become active participants in their health care. It is possible that health care providers in the hospital unit will be satisfied with the BMT Roadmap and its impact on the clinician-participant relationship.

Conclusions

This protocol offers a framework for design and analysis of personalized health IT systems to increase patient and caregiver engagement in acute care, hospital-based contexts.

Acknowledgments

The development of the BMT Roadmap was IRB approved (HUM00093082: Personalized Engagement Tool for Pediatric BMT Patients and Caregivers). The clinical trial, registered at ClinicalTrials.gov as NCT02409121, is pending IRB approval.

Conflicts of Interest

None declared.

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Abbreviations

BMT: blood and marrow transplantation
CHCR: Center for Health Communications Research
HCT: hematopoietic cell transplantation
EHR: electronic health record
IT: information technology
MCIT: medical center information technology
PAAQ: Parental Acceptance and Action Questionnaire
P-PAM: Parental-PAM

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Original Paper

Changes in Physical Activity and Psychological Variables Following a Web-Based Motivational Interviewing Intervention: Pilot Study

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Abstract

Background: Web-based interventions for enhancing physical activity participation are in demand for application in health care settings. Recent research suggests Web-based interventions that are based on motivational interviewing are effective to increase physical activity. It is unclear whether motivational interviewing can influence targeted psychological variables such as perceived readiness, willingness, and ability to participate in physical activity.

Objective: The aims of this study were to determine whether there were changes in physical activity and psychological variables associated with readiness, willingness, and perceived ability to participate in physical activity following completion of a novel Web-based intervention. The goal of the motivational interviewing–based intervention was to increase physical activity.

Methods: Twenty-three underactive or inactive urban dwelling adults were recruited at a medical office for participation in a 4-session Web-based intervention lasting approximately 15 minutes per week. Sessions were based on principles of motivational interviewing. Assessment of physical activity was conducted using pedometers immediately prior to intervention participation (pre) and immediately post intervention (post1). Self-report assessments of physical activity and psychological variables were conducted using online surveys at pre, post1, and again at one month following intervention participation (post2).

Results: Comparisons of pre and post1 pedometer recordings revealed significant increases in steps per day (t_{22}=2.09, P=.049). There were also significant changes in total physical activity energy expenditure per week (χ^2_2=8.4, P=.02) and in moderate intensity physical activity energy expenditure per week (χ^2_2=13.9, P<.001) over time following participation in the Web-based intervention. Significant changes in psychological variables following participation in the Web-based intervention included: (1) change in stage classification over time (χ^2_2=21.5, P<.001), where the percentage of participants classified in the action or maintenance stages of change in physical activity increased over time (pre=25% [6/24], post1=71% [17/24], post2=68% [15/22]); (2) decreases in self-reported decisional balance cons ($F_{2,42}$=12.76, P<.001); (3) increases in self-reported decisional balance pros ($F_{2,42}$=16.19, P<.001); (4) increases in physical activity enjoyment ($F_{2,20}$=3.85, P=.04); and (5) increases in self-efficacy ($F_{2,42}$=3.30, P=.047).

Conclusions: The Web-based intervention piloted in this study shows preliminary promise as a tool to promote physical activity in health care settings. Additional research is needed to test the effectiveness of motivational interviewing compared to a control condition and to refine content by considering mediation by psychological variables in a larger sample.

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KEYWORDS

motivational interviewing; physical activity; adults; Web-based; intervention; health care; psychology

Introduction

The United States is experiencing a public health crisis because most adults are not getting sufficient physical activity to promote health and prevent disease [1,2]. Interventions to increase physical activity that are effective and feasible to implement in health care settings are needed. One barrier to feasibility is that health care providers have limited face-to-face time with patients to discuss strategies for increasing physical activity [3]. Even when time allows, approaches such as direct advice-giving are often implemented and unfortunately are not likely to be effective [4]. To mitigate difficulties with lack of time, health care providers and patients alike may benefit from the availability of a Web-based intervention to which patients can be referred. While Web-based interventions are becoming more popular, there is not yet consensus on the specific framework of such interventions for increasing physical activity [5].

Motivational interviewing is one clinical technique [6] with preliminary support as a framework for Web-based delivery of interventions geared at increasing physical activity [7,8]. Motivational interviewing is thought to influence behavior change by enhancing a person's readiness, willingness, and perceived ability to engage in a particular behavior. The ready, willing, and able constructs of motivational interviewing map onto psychological variables. Psychological variables of interest specifically include readiness to change, decisional balance, enjoyment, and self-efficacy. Although readiness, willingness, and perceived ability are targeted during interventions that are based in motivational interviewing, research is needed to determine if this influences targeted psychological variables.

In summary, it has been established that interventions to increase physical activity are needed and that Web-based delivery may be desirable to address time limitations in health care settings. Although motivational interviewing-based interventions have shown preliminary support for increasing physical activity [4], there is limited research into whether Web-based motivational interviewing is effective for increasing physical activity when offered by health care providers. There is also limited research into the possible changes in psychological variables related to participating in motivational interviewing. Understanding whether interventions based on this method do in fact change psychological variables could provide valuable information for determining what specific content should be included in future iterations of interventions. Therefore, the aims of this study were to determine whether there were changes in physical activity as well as changes in psychological variables associated with readiness, willingness, and perceived ability following introduction to a novel Web-based intervention. The intervention was geared at increasing physical activity and was based on motivational interviewing. The specific aims of this study were to assess whether there were increases in physical activity and changes in targeted psychological variables following participation in the intervention.

Methods

Study Design

A repeated measures design was used to conduct a pilot assessment of whether there were changes in physical activity and psychological variables following participation in a Web-based intervention based on motivational interviewing. Data for both physical activity and psychological variables were gathered before intervention participation (pre), immediately after intervention participation (post1) and one month following intervention participation (post2). Links for intervention sessions were emailed to participants once per week for 4 weeks, and each session took approximately 15 minutes to complete.

Recruitment

Patients at an urban health care clinic in Milwaukee, Wisconsin, were recruited to participate. Flyers advertising weight management were placed at reception desks and circulated in waiting rooms. Patients who were interested provided contact information including personal and physician email addresses to the receptionists. Along with providing contact information, patients were asked whether they met the 2007 Centers for Disease Control and Prevention (US Department of Health and Human Services) guidelines for physical activity. Using the contact information provided, study staff sent email inquiries to the patients' physicians to obtain medical clearance for participation. In order to be eligible for participation, patients had to self-report insufficient physical activity, have a working email address, and have been cleared for participation by their medical doctor. Eligible and willing patients completed an online informed consent process before engaging in data collection and intervention activities.

Intervention

The intervention was novel and consisted of a Web-based program that was designed based on the principles of motivational interviewing. Each intervention session was self-guided and included prompts for the participant to engage in self-reflection on their physical activity behavior and related thoughts. The specific mechanism by which self-reflection was facilitated was that each session included a set of questions prompting the participant to generate a written response. All of the sessions were presented in black text with a white screen created using Qualtrics survey software. Textbox 1 shows an example of the series of questions included in the first session—questions which were based on motivational interviewing principles of building rapport, creating a discrepancy between current physical activity behaviors and goals, and addressing barriers to physical activity. Note that each question was followed by a text area for the participant to respond and a button to advance to the next screen.

Textbox 1. Examples of questions prompting participant self-reflection.

- Welcome! Glad you decided to participate! Your doctor talked to you about your physical activity. What are some of your concerns about how much physical activity you are getting?
- Those are certainly valid concerns. Thank you for sharing. Now that you have shared your concerns, let's consider some of your thoughts about physical activity. What are some reasons that you aren't getting as much activity as you or your doctor would like?
- Those are difficult challenges to overcome, and you've done a great job thinking this through. It really is hard to get as much activity as is recommended. Describe, if you would, a time in the past when you were more active or activities that you were able to do and how those challenges fit in.

Notice that the tone of the questions was such that the participant was greeted with validation and acceptance, consistent with the tone of motivational interviewing. Responses were prompted with the goal of guiding the participant's thinking in the direction of feeling ready, willing, and able to engage in physical activity. One deviation from traditional motivational interviewing was that the questions were not individually tailored. That said, after the first session participants were sent a personalized email message thanking them for participation and reflecting on their responses. This email message was the only tailored component of the intervention and was implemented to facilitate participant retention.

Sessions two through four followed the same format and delivery style as the first session. During the second session, content included self-reflection on feedback about physical activity and was geared toward eliciting a commitment to change physical activity behaviors. The third session involved creation of a change plan and was geared at strengthening the commitment to change. During the fourth and final session, efforts to change were affirmed, and commitment to change was further reinforced.

Measures

Overview

Demographic information was gathered using an online self-report inventory at pre. Online self-report instruments were also used to gather subjective reports of physical activity and psychological variables at pre, post1, and post2. Pedometer recordings were gathered by the participant and emailed to the researchers at pre and post1. To avoid the potential confounding influences of having participants monitor steps, participants were instructed not wear their pedometers or record steps during the intervention phase.

Physical Activity

Assessment of physical activity was conducted using both pedometers and self-report surveys. Using pedometers is recommended to accurately capture total amounts of physical activity among adults, given the likelihood of underestimation of physical activity during self-reports [9]. As such, Yamax SW-200 pedometers were used to measure steps taken per day. Total steps over the course of 7 days was recorded with the pedometer worn at the hip. Each participant was given instructions for recording and emailing daily step-counts to the principal investigator. Pedometer data were compiled by tabulating the total number of steps recorded by the participant each day for 7 days. Yamax pedometers have previously demonstrated a high degree of reliability given estimates of 100% consistency in counting steps when compared to hand-tallied observed step-counts [10].

For the self-report tool, the International Physical Activity Questionnaire (IPAQ) short form for self-administration was used to assess the number of minutes spent participating in physical activity in the past 7 days [11]. The participants were asked to report number of days per week and hours and minutes of engagement in behaviors classified as vigorous, moderate, walking, and sitting. Total physical activity metabolic equivalent of task minutes (METs) per week were calculated in accordance with IPAQ scoring guidelines by summing the following: (1) 3.3 × walking minutes × walking days, (2) 4.0 × moderate intensity activity minutes × moderate days, and (3) 8.0 × vigorous intensity activity minutes × vigorous intensity days. The results of prior research indicate that the IPAQ has sufficient test-retest reliability (r=.66-.88, 95% CI 0.73-0.77) and moderate criterion validity against accelerometers when measuring moderate intensity physical activity among adults (r=.46-.81, 95% CI 0.23-0.36) [11].

Psychological Variables

Readiness

Readiness to engage in physical activity was assessed by examining participant stage classification using the Exercise Stages of Change Questionnaire [12]. Individuals were categorized in stages precontemplation, contemplation, preparation, action, or maintenance, where higher stages such as action and maintenance reflect more engagement in physical activity compared to lower stages. The Exercise Stages of Change Questionnaire showed modest significant correlations with step recordings at pre (r=.38, P=.02) and post1 (r=.47, P=.03). The Exercise Stages of Change Questionnaire has previously shown high test-retest reliability among adult men and women over a two-week period with a kappa index of .78 [13].

Willingness

Willingness to engage in physical activity was assessed by examining both decisional balance related to physical activity and physical activity enjoyment. A decisional balance questionnaire [14] was used to ask participants to rate the importance of statements pertaining to making decisions about whether to engage physical activity. Greater endorsement of exercise pros and lower endorsement of exercise cons would suggest greater likelihood of engaging in physical activity. In this study, the decisional balance questionnaire demonstrated internal consistency reliabilities that were acceptable to high for pros at pre (r=.62), post1 (r=.39), and post2 (r=.94) as well as cons at pre (r=.72), post1 (r=.81), and post2 (r=.76).

Higher scores on the Physical Activity Enjoyment Scale (PACES) [15] indicated greater enjoyment. Internal consistency ratings were high in the study at pre (r=.96), post1 (r=.93), and post2 (r=.94), and the scale previously demonstrated discriminant validity when correlated with boredom proneness among adults during exercise (r=−.30, P<.05) among adults [16].

Perceived Ability

Perceived ability to engage in physical activity was assessed using a self-efficacy questionnaire [17] where higher scores indicated greater perceived ability. Internal consistency ratings were good in the study at pre (r=.74), post1 (r=.81), and post2 (r=.74).

Statistical Analysis

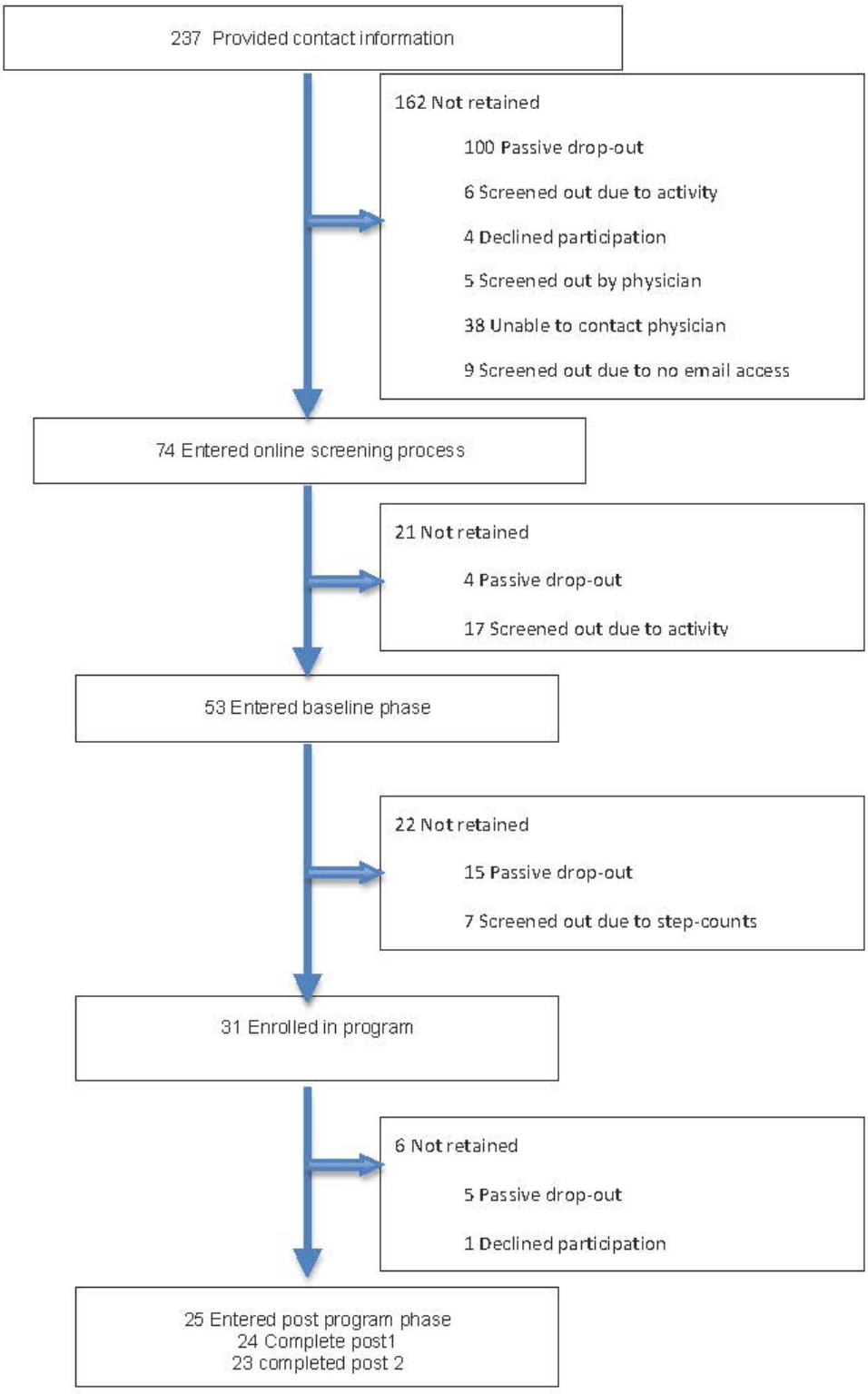
Step-count data were normally distributed and therefore paired samples t tests were completed to compare average steps per day from pre to post1. Due to positive skew of IPAQ data, Friedman nonparametric tests with post hoc Wilcoxon analyses were conducted to assess changes in self-reported physical activity over time. Friedman and Wilcoxon signed-rank follow-up tests were also conducted to assess changes in stage classification over time. Since decisional balance and self-efficacy data were normally distributed, a series of repeated measures analyses of variance (ANOVAs) was completed to assess for change over time. Enjoyment data violated the sphericity assumption; therefore the multivariate test results including Wilks lambda values were interpreted from the repeated measures ANOVAs. Analyses were conducted using SPSS version 22.0 statistical software (IBM Corporation).

Results

Participant Characteristics

Among the 237 persons who responded to the recruitment flyer, 31 enrolled in the intervention, and ultimately 23 participants (87%, 20/23, female; mean age 46, SD 11.0) completed all assessments and intervention sessions. See Figure 1 for a flow chart of recruitment, enrollment, and retention. The cohort of participants who completed the study was racially diverse, with 44% (10/23) of the participants identifying as other than white. The majority of the participants 96% (22/23) reported being overweight or obese.

Figure 1. Recruitment, enrollment, and retention.



Changes in Physical Activity

Significant increases were found for average daily steps from 5252.99 (SD 2412.66) at pre to 6425.05 (SD 2783.73) at post1 (t_{22}=2.09 P=.049), total weekly energy expenditure was 1918.33 METS at pre (SD 2755.11) to 3457.12 METS at post2 (SD 5670.63) (χ^2_2=8.4 P=.02), and moderate weekly energy expenditure 218.00 at pre (SD 417.15) to 1091.00 (SD 2589.88) at post2 (χ^2_2=13.9, P<.001).

Changes in Psychological Variables

Stage classification percentages changed significantly over time (χ^2_2 =21.5, *P*<.001) and are detailed in Table 1. Mean ranks increased from pre (1.39) to post1 (2.23) and post2 (2.39). Follow-up testing revealed significant changes from pre to post1 (*z*=3.02 *P*=.00 and pre to post2 (*z*=3.37, *P*<.001) but not post1 to post2 (*z*=0.79, *P*=.43). Cross-tabulation results (see Table 1) further reveal that while there were no participants in the precontemplation stage at any point, the percentage of participants in the contemplation stage was lower at post1 and post2 compared to pre, whereas more participants were in the action and maintenance phases at post1 and post2 compared to pre.

Table 1. Percentage of participants in stage classifications pre, post1, and post2.

	Pre (n=24) n (%)	Post1 (n=24) n (%)	Post2 (n=22) n (%)
Contemplation	15(63)	7 (29)	5 (23)
Preparation	3 (13)	0	2 (9)
Action	5 (21)	13 (54)	8 (36)
Maintenance	1 (4)	4 (17)	7 (32)

Sources of variance, *F* ratios, and significance values for the repeated measures ANOVA detailing significant changes over time in psychological variables associated with willingness and perceived ability are provided in Table 2. There were significant changes in directions for decisional balance pros, decisional balance cons, physical activity enjoyment, and self-efficacy. Notably, these changes were in the expected direction given that relevant to physical activity, participants self-reported increases in decisional balance, enjoyment, and self-efficacy over time.

Table 2. Repeated measures ANOVA source table for psychological variables over time.

Source		Degrees of freedom	Sum of squares	Mean sum of squares	F statistic	P value
Decisional balance, pros (n=22)						
	Between groups	2	654.21	327.11	16.19	<.001
	Within groups	42	848.46	20.20	—	—
Decisional balance, cons (n=22)						
	Between groups	2	357.21	178.61	12.76	<.001
	Within groups	42	588.12	14.00	—	—
PACES (n=22) [a]						
	Between groups	2	—	—	3.85	.04
	Within groups	20	—	—	—	—
Self-efficacy (n=21)						
	Between groups	2	161.85	80.92	3.30	.047
	Within groups	42	1030.82	24.54	—	—

[a]Wilks lambda values reported due to sphericity assumption violation.

Discussion

Principal Findings

Overall, results of the pilot study were promising regarding effectiveness of the Web-based intervention tested. There were changes in physical activity as well as changes in psychological variables associated with readiness, willingness, and perceived ability to participate following participation in the motivational interviewing–based intervention. Changes were evident based on significant increases in both step-counts from pre to post1 and self-reported moderate and total weekly physical activity. Changes in psychological variables over time included significant increases in physical activity–related stage classification, decisional balance, enjoyment, and self-efficacy. One plausible explanation for the observed increases in physical activity is that changes in psychological variables (ie, readiness, willingness, perceived ability) contributed to enhanced participation in physical activity. While the changes in psychological variables may have come about due to the direct impact of the motivational interviewing–based intervention, this assertion cannot be fully supported without further research. Nonetheless, preliminary findings are encouraging.

Results regarding physical activity and psychological variables are particularly meaningful given the strengths of the current

study. With regard to assessment of physical activity, the use of both pedometers and self-report surveys builds confidence that observed changes were valid [10]. Further, although the sample size was small, the sample was racially diverse and likely representative of the population to which the intervention would apply. That is, the sample included adult patients at an urban health care clinic who were not engaging in sufficient physical activity. Although patients who were overweight or obese were not explicitly targeted for recruitment, the sample comprised individuals who could benefit from engaging in additional physical activity due to being classified as overweight or obese.

In addition to testing effectiveness using triangulated methods to assess physical activity in a relevant sample, the study examined changes in psychological variables (variables that have not been included in prior research). That is, research on changes in psychological variables targeted during interventions that are Web-based and are based on motivational interviewing was not previously available. Acknowledging changes in targeted psychological variables is critical for understanding the mechanisms by which the intervention is likely to impact physical activity.

Limitations

The limitations discussed should be taken into consideration when interpreting the results of this study and when planning future research. Due to lack of a control group, causal impact of the intervention cannot be determined. Nonetheless, because changes in physical activity and psychological variables at post1 and post2 are in the expected direction and are in agreement over time, the pilot results serve as a basis for supporting further research. To address this limitation, future research efforts should include a control condition.

Recruitment of a larger sample is necessary to allow sufficient statistical power for comparing the intervention condition to a control condition. Sample characteristics in the current study also reveal greater participation by women who were overweight or obese. In order to determine whether selective drop-out by men or persons of healthy body weight occurred, systematic investigation of those individuals who enrolled but did not follow through with intervention participation may be warranted.

Interpretation of stage classification outcomes is complicated by potential validity issues in the study. Validity concerns specifically pertain to questionable accuracy of stage classification given that more participants self-reported advancing to the maintenance stage (which necessitates 6 months of behavior change) at post2 than is logical or possible when taking into account that only 2 months had elapsed since pre. Because of questionable validity of the stage of change outcomes, continued triangulation with other sources of physical activity data is recommended.

Conclusions

Results are consistent with previous research, in that they provide further support that Web-based interventions based in motivational interviewing show promise for increasing physical activity participation [7,8]. The Web-based motivational interviewing intervention piloted in the study demonstrates preliminary evidence of effectiveness for enhancing physical activity in a health care setting. The mechanisms by which the motivational interviewing-based interventions impact physical activity may be through changes in targeted psychological variables. Collectively, these findings support the need for additional investigation into the effectiveness and mechanisms of the intervention that was piloted. Specifically, a meditational model should be tested to determine if the intervention directly influences psychological variables and whether changes in psychological variables indirectly contribute to increases in physical activity.

The intervention tested was created to meet the demands of time-limited physicians and their patients. The findings indicate that this intervention may in fact be valuable for health care delivery given that the intervention was effective for increasing physical activity in a health care setting. Benefits of the intervention are that it was very brief and the delivery format required limited involvement of health care professionals. Specifically, the intervention sessions required approximately 15 minutes of participation per week for a total of four weeks, and delivery of the intervention required only emailing links to the participants each week, with one follow-up email provided after the first session. The tasks associated with intervention delivery could be conducted by an individual with less training than a physician, thereby alleviating burden on physician time and still providing a necessary resource for patients who are not getting sufficient physical activity for health and well-being.

Conflicts of Interest

None declared.

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Abbreviations

IPAQ: International Physical Activity Questionnaire
MET: metabolic equivalent of task
PACES: Physical Activity Enjoyment Scale
Pre: prior to intervention participation
Post1: immediately following intervention participation
Post2: one month following intervention participation

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Corrigenda and Addenda

Addendum to: The Mobile Insulin Titration Intervention (MITI) for Insulin Glargine Titration in an Urban, Low-Income Population: Randomized Controlled Trial Protocol

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The authors of "The Mobile Insulin Titration Intervention (MITI) for Insulin Glargine Titration in an Urban, Low-Income Population: Randomized Controlled Trial Protocol" (JMIR Res Protoc 2015;4[1]:e31) are adding their insulin glargine titration algorithm to this protocol paper. The authors feel that readers may be interested in seeing the algorithm used in the trial. The algorithm was added as Multimedia Appendix 2 to the paper and the article was resubmitted to Pubmed Central.

Multimedia Appendix 2

Insulin titration algorithm.

[PDF File (Adobe PDF File), 4KB - resprot_v4i4e138_app2.pdf]

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