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Comparison of Two Theory-Based, Fully Automated Telephone Interventions Designed to Maintain Dietary Change in Healthy Adults: Study Protocol of a Three-Arm Randomized Controlled Trial

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Abstract

Background: Health behavior change interventions have focused on obtaining short-term intervention effects; few studies have evaluated mid-term and long-term outcomes, and even fewer have evaluated interventions that are designed to maintain and enhance initial intervention effects. Moreover, behavior theory has not been developed for maintenance or applied to maintenance intervention design to the degree that it has for behavior change initiation.

Objective: The objective of this paper is to describe a study that compared two theory-based interventions (social cognitive theory [SCT] vs goal systems theory [GST]) designed to maintain previously achieved improvements in fruit and vegetable (F&V) consumption.

Methods: The interventions used tailored, interactive conversations delivered by a fully automated telephony system (Telephone-Linked Care [TLC]) over a 6-month period. TLC maintenance intervention based on SCT used a skills-based approach to build self-efficacy. It assessed confidence in and barriers to eating F&V, provided feedback on how to overcome barriers, plan ahead, and set goals. The TLC maintenance intervention based on GST used a cognitive-based approach. Conversations trained participants in goal management to help them integrate their newly acquired dietary behavior into their hierarchical system of goals. Content included goal facilitation, conflict, shielding, and redundancy, and reflection on personal goals and priorities. To evaluate and compare the two approaches, a sample of adults whose F&V consumption was below public health goal levels were recruited from a large urban area to participate in a fully automated telephony intervention (TLC-EAT) for 3-6 months. Participants who increase their daily intake of F&V by ≥1 serving/day will be eligible for the three-arm randomized controlled trial. A sample of 405 participants will be randomized to one of three arms: (1) an assessment-only control, (2) TLC-SCT, and (3) TLC-GST. The maintenance interventions are 6 months. All 405 participants who qualify for the trial will complete surveys administered by blinded interviewers at baseline (randomization), 6, 12, 18, and 24 months.

Results: Data analysis is not yet complete, but we hypothesize that (1) TLC-GST > TLC-SCT > control at all follow-up time points for F&V consumption, and (2) intervention effects will be mediated by the theoretical constructs (eg, self-efficacy, goal pursuit, conflict, shielding, and facilitation).

Conclusions: This study used a novel study design to initiate and then promote the maintenance of dietary behavior change through the use of an evidence-based fully automated telephony intervention. After the first 6 months (the acquisition phase), we will examine whether two telephony interventions built using different underlying behavioral theories were more successful than an assessment-only control group in helping participants maintain their newly acquired health behavior change.

Trial Registration: Clinicaltrials.gov NCT00148525; http://clinicaltrials.gov/ct2/show/NCT00148525 (Archived by Webcite at http://www.webcitation.org/6TiRriJOs).

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KEYWORDS

maintenance; social cognitive theory; goals; fruit; vegetable; diet; telephone; health information systems

Introduction

Lifestyle behaviors, including smoking cessation, prevention of overweight and obesity, physical activity, and healthful diets, are recommended for health promotion and disease prevention across a wide range of chronic conditions including cardiovascular disease, diabetes, and cancer [1]. Epidemiological studies have examined both overall eating patterns and intake of individual foods and nutrients for their effects on overall mortality and specific diseases [2-4]. While research examining relationships between diet and disease is complex due to issues of measurement, self-reporting bias, integration of foods within the total diet, confounding, among other issues, a large compilation of research supported the rationale that particular diet behaviors can affect diet-related cancer risk, including probable evidence of decreased risk with intake of foods high in dietary fiber (colorectal cancer) and fruits and vegetables (mouth, pharynx, larynx, esophagus, and stomach cancers) [3].

Surveillance of eating patterns in the United States indicates that the majority of the population does not meet recommendations for multiple dietary components, including fruits and vegetables (F&V), which are consumed at approximately half of recommended levels [5]. A comprehensive review of 45 studies [6] provides evidence that specific dietary interventions can lead to modest effects on improving diet. For interventions that targeted F&V intake, the average amount of change has ranged from about a 0.5 serving to slightly over one serving per day increase [6-13]. Given a large cohort study that found a 53% higher mortality rate among those who consume no fruits and vegetables compared with those who eat 5 servings a day as well as a dose-response relationship between increasing levels of F&V intake and overall mortality [2], it is expected that even modest increases in F&V intake would lead to beneficial mortality outcomes.

In recent years, multiple commentators have called for the need to sustain short-term health behavioral intervention effects by studying intervention effects at the end of the intervention period as well as long-term follow-up after the intervention concluded. Gaining a better understanding of behavior change maintenance was highlighted by Kumanyika et al more than 10 years ago [14] and more recently by the Health Maintenance Consortium in 2010 [15-18]. A systematic review of the maintenance of dietary change found that while 90% of the trials reported significant outcomes at the end of the intervention, 35% reported diet and/or physical activity significant outcomes at least 3 months after the intervention ended. However, of the seven diet trials that reported long-term outcomes (range 3-12 months following the conclusion of the intervention), there were promising effects on dietary outcomes, with six trials reporting significant between group differences (ie, maintenance) of at least one dietary outcome [19]. Additional dietary interventions have been published since the time of that review, which reported significant maintenance of effects ranging from 6 months to a year after the end of the diet intervention [20-23]. For example, a telephone-based intervention using the Get Healthy Information and Coaching Service in Australia demonstrated that a 6-month intervention followed by 6 months of no contact, participants maintained their increase in fruit intake and decreases in weight, waist circumference, and body mass index (BMI) [22]. In another telephone-based intervention, intervention effects for percent calories from total fat and saturated fat, fiber, and fruit were maintained after a 12-month intervention followed by 6 months of no contact [21]. Interestingly, for both telephone-delivered interventions, maintenance of effects for vegetable intake was not sustained during the periods of no intervention contact despite initial improvements immediately following the interventions.

One aspect limiting the further development of the science of behavior change maintenance is the lack of theories that have been developed to specifically address maintenance versus initiation of a health behavior change. Some experts suggest that a new theoretical approach may be needed to understand the maintenance of behavior changes and how to design interventions [24,25]. Rothman proposes that maintenance of a behavior change may involve different cognitive processes than initiation of behavior change [26]. Theories that have been used to inform behavioral interventions (eg, social cognitive theory) may fall short in that they use the same processes to initiate and to maintain a behavior change.

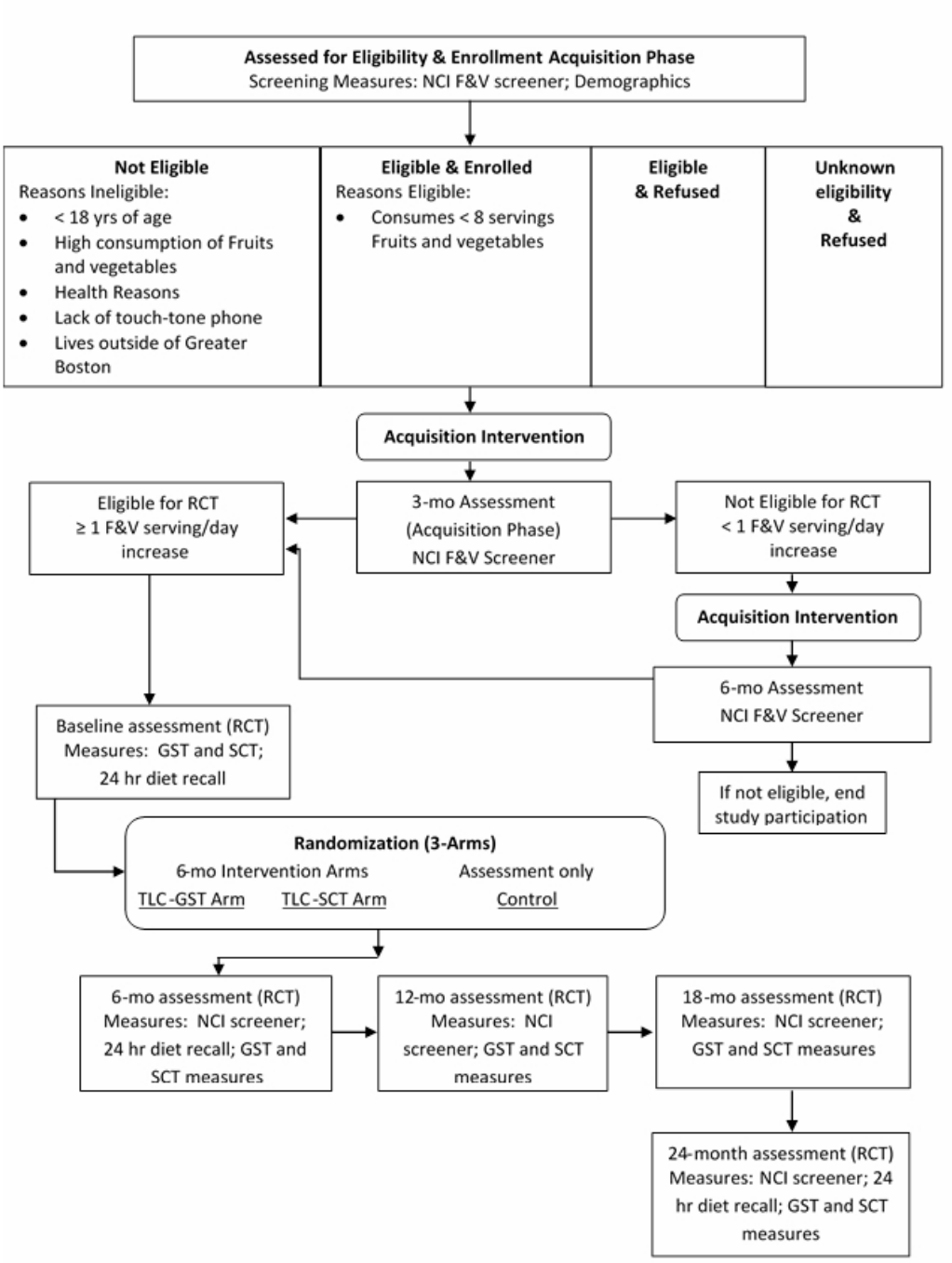
The present study conceptualizes maintenance as a distinct phase of the health behavior change process and proposes Goal Systems Theory (GST) [27-30] as a possible theory to inform the design of a maintenance intervention to help individuals manage their newly acquired behavior (increased consumption of F&V) following the acquisition of this behavior. This novel approach will be compared to a widely accepted, evidence-based framework that guides the design and evaluation of many dietary interventions—social cognitive theory (SCT) [31]. This paper describes the research design used to evaluate two theory-based interventions designed specifically to assist with the maintenance of a newly acquired dietary behavior (F&V consumption), one intervention guided by SCT and the other guided by GST.

Methods

Overview

The design for this study includes two phases to examine the maintenance of a dietary behavior change in adults. Figure 1 provides an overview of the study flow and measurement time points. In the first phase (acquisition), an intervention was used to produce a change in F&V consumption in order to study, in a second phase (maintenance), if an intervention targeting maintenance could sustain that change. All interventions were delivered using a fully automated telephony system that used an interactive voice response (IVR) system to generate speech that emulated counseling by a trained behavioral counselor combined with a speech recognition system to understand what the participant said. To perform the study, a sample of healthy adults who consumed less than the recommended level of F&V (ie, ≤ 5 servings/day) were recruited. During the acquisition phase, they were given a tested dietary telephony intervention for up to 6 months, called Telephone-Linked Care (TLC)-EAT, which has been shown in previous studies to have positive effects on initiating dietary improvements, including increased F&V consumption [32,33]. Because both prior studies found an increase of approximately one serving of F&V for the TLC-EAT intervention group, it was hypothesized that this effect would occur in a new sample of participants. F&V intake was examined at enrollment and after 3 months of the acquisition intervention. Participants who increased their consumption of F&V by at least one serving/day became eligible for the second phase (maintenance). Participants who did not show a one-serving increase were allowed to participate for an additional 3 months only. Those who achieved a one-serving increase during the acquisition study were eligible to participate in the second phase (maintenance, randomized controlled trial). Participants who entered the second (maintenance) phase of the study were blinded to the inclusion criterion (eg, an increase of >1 serving/day of F&V). Participants in the maintenance phase were randomized to one of three groups, receiving one of two theoretically based TLC interventions targeting maintenance of behavior (TLC-Maintenance GST or TLC-Maintenance SCT) or to the control group (an assessment only without an intervention). All participants in the randomized controlled trial (RCT) were assessed every 6 months for a total of 2 years post randomization (baseline). The study protocol received full board review and approval by the Boston University School of Medicine Institutional Review Board (NCT00148525). See Multimedia Appendix 1 for the CONSORT-EHEALTH checklist [34].

Figure 1. Flow diagram of the study.



Participants

The pool of possible participants was randomly selected from the voter registration lists from the Boston metropolitan area. Participants were required to be ≥18 years of age, live in the Boston area, have access to a touch-tone telephone, and be generally healthy. Participants were also required to be "under-consumers" of F&V as defined by eating fewer than 5 servings/day. However, in the first 2 months of recruitment, a large proportion (62%) reported consuming more than 5 servings/day using the National Cancer Institute F&V Screener (NCI FVS) [35]. Given that this measure may overestimate the servings of F&V [36-38], the inclusion criterion was modified to "consuming less than eight F&V servings/day" in order to correct for the bias in the NCI FVS. This modification decreased the percentage of screened individuals who were not eligible (39%) yet still captured adults who were not meeting a public health goal level, for example, the 2005 dietary guidelines of 9 servings/day of F&V [39-41]. Participants were not eligible if they were pregnant or if they had a recent health event such as a diagnosis of cancer, myocardial infarction, kidney disease, eating disorder, or were prescribed a special diet.

Procedures

The University of Rhode Island Survey Research Center performed study recruitment, assessment, and randomization. The Center randomly selected names from a voter registration list and mailed a letter inviting them to participate in the study.

The letter was followed by a phone call from the Center about one week later where the interviewer described the study, screened for eligibility (see Participant section above for eligibility criteria), obtained verbal consent, and administered study instruments. Research assistants at Boston University School of Medicine, where the intervention (ie, TLC-EAT) was hosted, sent participants an enrollment packet in the mail, which contained a welcome letter, consent form for their records, and a manual and personal password for the healthy eating acquisition intervention (ie, TLC-EAT). A week after enrollment, these research assistants called participants to train them on how to use TLC-EAT. Participants did a practice call with the research assistant on the line. Those who completed the training were transferred to the first intervention call (the "training call") with TLC-EAT. Thereafter, all TLC calls were outbound calls initiated by the automated system, which called participants at the time initially entered into the automated scheduling system during the training call. Participants were asked to complete one TLC-EAT call per week for 12 weeks with the option of rescheduling any of the incoming TLC calls or initiating calls to TLC if preferred. After 12 weeks, F&V consumption was reassessed with the NCI FVS to determine eligibility for the maintenance phase of the trial. Participants who increased by one serving of fruit and/or vegetable were eligible to be randomized to one of three groups for the RCT. Random allocation to group assignment was generated by the SRC's computer program that used urn randomization protocols to balance groups by gender. Those who did not increase by one serving after 3 months were invited to continue with the acquisition intervention, TLC-EAT, for an additional 3 months and then were assessed again for eligibility into the RCT. Those who qualified and agreed to continue in the RCT were assessed at baseline (randomization), 6, 12, 18 and 24 months post baseline. A random sample of those who qualified for the RCT were invited to complete a blood draw at the university's General Clinical Research Center at baseline only. Assessments during the RCT included the NCI FVS, 24-hour dietary recalls, and psychosocial measures. Participants received US $20 for completing a survey at baseline, 6-month, 12-month, 18-month, and 24-month time points, and $50 for the in-person, fasting blood draw.

Intervention

Overview

All interventions were delivered using a fully automated telephony system, TLC, which speaks to participants using computer-controlled, pre-recorded human speech, and the participant selects among pre-determined options to respond to the computer by either pressing keys on the telephone keypad or selecting an option by speaking into the phone [42]. TLC systems deliver an individualized intervention that mimics a conversation between a counselor and client.

Dietary Acquisition Intervention

The acquisition phase was delivered by TLC-EAT, designed to improve general diet quality by broadly targeting important nutrients in the diet, such as saturated fat and fiber, and by targeting food groups (eg, fruits, vegetables, whole grains), and by encouraging related dietary behaviors such as avoiding fried foods. See Multimedia Appendix 2, Table 1, for topics and previously published studies for details [32,33]. However, two modifications were made to TLC-EAT for the present study. The participants did not receive the two mailed printed reports received in previous trials. The second modification was that the present study used an "outbound" system (TLC contacts participant) to call participants on a weekly schedule instead of an "inbound" calling system (participant contacts TLC) used in the previous trials.

Maintenance Interventions

Two novel, theory-based interventions were used to promote the maintenance of dietary change with an emphasis on fruits and vegetables. Participants randomized to one of the two intervention arms of the RCT received ten automated TLC calls over the course of 6 months with the frequency of contacts reduced over time. These participants received one call per week in the first month (four calls in month one), one call every other week in the second month (two calls in month two), and one call per month for the remaining 4 months (one call in months three, four, five, and six). Calls were 10-15 minutes in duration. The first call after randomization was the same for both intervention groups and consisted of feedback on F&V, whole grains, low-fat dairy, and saturated and trans fats (see Table 2 in Multimedia Appendix 2). This call provided feedback for each of the food groups using data obtained from the Prime Screen [43], a screening instrument administered at the screening and randomization time points and used for intervention purposes only. Feedback consisted of progress made since the start of the acquisition intervention and comparisons to dietary recommendations. The remaining nine calls were different in content for the two intervention groups but not in duration (eg, 10-15 minutes).

Goal Systems Theory

GST posits that goals, as cognitive constructs, are mentally represented and organized and that this organization may help determine how goals are chosen and pursued [27-30]. This mental organization of goals into a system or structure assumes a hierarchy with primary goals and sub-goals. Primary goals are more abstract in nature yet have a large number of concrete means that represent specific behaviors used to attain that goal. GST also assumes that goals are linked to each other, and these inter-goal connections may play an important role in goal choice, goal pursuit, and goal attainment.

Maintaining a goal may depend on the characteristics of the goal and where that goal is within the goal system. A goal's characteristics can include its perceived difficulty and value and its connection to other goals. Goals that are perceived as facilitating another goal are more likely to be pursued than goals that compete or conflict. Goals can be perceived as redundant or substitutable with other goals in one's system because they fulfill the same underlying need or desire of another goal (eg, goal of exercising regularly to achieve health vs goal of healthful diet to achieve health). GST posits that the maintenance of goal pursuit will decrease the more the individual perceives the goal as substitutable or redundant with other pursuits. Thus, the greater the degree to which a goal is perceived to facilitate other goals while the less it is perceived to be redundant with these

goals, the greater the likelihood its pursuit will be maintained. GST also suggests that the maintenance of pursuing a goal may be enhanced to the degree to which an individual integrates current pursuits with his or her other pursuits. GST-related research has found that integrating goals is associated with positive health outcomes [27,44-46].

Telephone-Linked Care Maintenance–Goal Systems Theory

GST was used to develop a TLC program that would query each TLC-Maintenance Goal Systems (TLC-GST) participant on how the person manages a diet goal (maintaining increased consumption of F&V) and other life goals. The general thesis is that maintenance of behavior change fails because of reasons outside of the specific behavioral domain, namely competition from other life goals. TLC-GST's purpose was to train the participant in goal management techniques to help them maintain dietary changes. The four domains of goal management targeted in TLC-GST were (1) finding ways to reduce conflict between competing goals (ie, goal conflict reduction) [27], (2) applying strategies that use other goals to help attain diet goals (ie, cross goal facilitation [47]), (3) finding ways to protect or shield the targeted goal from other competing goals (ie, goal shielding [30]), and (4) enabling the individual to maintain the resources that are necessary for achievement of the targeted goal (eg, maintenance of increased F&V consumption) (ie, facilitating goal maintenance [48]).

TLC-GST used a cognitive-based approach in which participants were asked to think and reflect instead of the traditional skills-based approach generally used in SCT interventions. As described above, the first maintenance call was the same for both SCT and GST. The GST intervention began after this first call. The general structure of a TLC-GST counseling call began with a greeting, followed by counseling on a GST-related topic (see the section on Goal System Theory above), homework of suggested exercises, and a closing that included a reminder about the next call. In each of these calls, TLC-GST reminded the participants of their top four ranked life goals collected at randomization (see Table 3 in Multimedia Appendix 2) and asked them to select one to discuss on the call. The content of the conversation focused on a life goal that the participant chose, its attributes, and its relation to dietary (eg, F&V) goals. As mentioned earlier, homework was assigned at the end of each call (eg, "I'd like you to write down 3 of your larger goals. Then, think about how healthy eating can help you to meet those larger goals"); however, the strategy of goal setting, which was used in TLC-SCT, was not part of the TLC-GST intervention. See Table 2 in Multimedia Appendix 2 for more details on call structure and content.

Social Cognitive Theory

Within SCT, there are five constructs that are relevant to health interventions, namely knowledge, self-efficacy, outcome expectation, goal formation, and social-environmental factors [31]. The present maintenance study focused on increasing self-efficacy. Self-efficacy is related to whether a person will attempt a task and also to how long a person will persevere. Self-efficacy can be increased using strategies such as providing specific feedback, positive reinforcement, encouraging small steps towards a goal and goal setting. Evidence suggests that interventions designed to increase self-efficacy improve adherence to health behaviors [49-51], and it may be one of the strongest mediators of behavior change [52]. A health behavior model that draws from social cognitive theory, targets self-efficacy as a mediator of behavior, and focuses on maintaining a behavior change is the Relapse Prevention Model (RPM) [53]. A key strategy with RPM is the identification and anticipation of situations that can lead to relapse back to unhealthy behaviors. Planning ahead for these risky situations can increase the probability that an individual will be successful when faced with this challenge. When small successes are experienced, self-efficacy increases. Thus, in the present study the strategies of planning ahead to overcome or avoid risky situations were used to help maintain a newly acquired behavior and increase self-efficacy.

Telephone-Linked Care Maintenance–Social Cognitive Theory

The intervention was designed to assist with the maintenance of diet changes using intervention components to build self-efficacy. TLC-SCT was developed as an extension of the acquisition study intervention (TLC-EAT) with a focus on maintaining healthy eating rather than obtaining it. RPM [53] was used to inform what strategies should be included in a maintenance intervention, although the overarching goal for the ten calls was to remind participants about the skills they learned previously and continue to build knowledge and skills. The main skills included goal setting, identifying barriers to healthy eating, anticipating and planning ahead for situations that lead to unhealthy eating, and rewarding oneself for reaching goals. TLC addressed knowledge within the context of maintenance and provided specific, positive feedback on behaviors and goals obtained, an important component of building self-efficacy and mastery.

The dietary content for the ten TLC-SCT maintenance calls focused on F&V as well as the food groups targeted in TLC-EAT. The food group topics mirrored that of TLC-EAT because the initial changes to the individual's F&V happened within the context of these food groups. Different from TLC-EAT, fruit and/or vegetable intake and goals were emphasized on every TLC-SCT call in addition to the food group topic. As mentioned previously, the first call for TLC-GST and TLC-SCT were the same. After the first call, the general structure of a call was (1) follow up on goals set previously, (2) assess intake of a food group and confidence to improve or maintain intake, (3) participant selects a barrier that impedes healthy eating and receives strategy for overcoming that barrier, (4) participant selects situation that tends to lead to relapse and hears tips for planning ahead, (5) participant given the option to set a goal for the main food group discussed and an option to set an additional fruit or vegetable goal, and (6) end with a take-away message about the call.

Measures

Overview

Assessments for the acquisition phase included self-report questionnaires administered at the enrollment call, and 3 months

and 6 months later. Measures included demographics, self-reported weight and height, and the NCI FVS. Additional measures were added to the maintenance phase (RCT) to more accurately estimate F&V outcomes and to examine change in theoretical constructs from SCT and GST. Baseline (randomization), 6 months, and 24 months were considered the primary time points for the RCT. Assessments at 12 and 18 months post baseline were considered secondary. A subsample of participants who were eligible for the RCT were randomly selected and invited to have a blood draw at baseline (randomization) to assess serum carotenoids. Other than the in-person blood draw, all assessments were completed over the telephone by survey research staff who were blinded to condition.

Fruit and Vegetable Intake

The primary outcome measure was F&V, and it was assessed two ways: brief screeners and 24-hour dietary recalls. The NCI FVS [35] was considered the primary screener and was administered at all assessment time points. The screener assesses self-reported frequency of consumption in the last month and the portion size for 10 items that included 100% fruit juice, fruit, lettuce salad, french fries or fried potatoes, white potatoes (not fried), cooked dried beans, other vegetables, tomato sauce, vegetable soup, and mixtures that include vegetables. Although all questions were asked, fried potato consumption and mixtures that include vegetables were not included in the summary scores or analyses. Validity of the FVS compared to true intake ranges from r=.66 for men to r=.51 for women [35].

For the primary assessment time points for the RCT (baseline, 6, and 24 months), the primary assessment of F&V was the NCI Method [54,55]. This method requires a food frequency assessment such as the NCI FVS and two 24-hour dietary recalls. The recalls were administered over the telephone by the Nutrition Epidemiology core at University of North Carolina, Chapel Hill, using the latest version of the Nutrition Data System for Research (NDSR) software. A nutritionist trained to conduct the telephone recalls used a standard introduction script and a multiple-pass approach interview methodology for the recall. The foods, beverages, preparation methods, amounts, and recipes reported by the participant were entered by an interviewer into the NDSR software to obtain an estimate of nutrient intake. The NDSR 2008 database was used for this study and contained over 18,000 foods, 8000 brand name products, many ethnic foods, supplements, and vitamins. The NDSR calculated food group serving count system was used to assess F&V.

Maintenance of F&V consumption was defined in this study as sustaining a one-serving/day increase in F&V achieved during the acquisition phase. Participants who increased their consumption by more than one serving during the acquisition phase need only maintain a one-serving gain to be considered as achieving maintenance. Maintenance (sustaining at least one serving/day) will be assessed at each time point, separately.

Carotenoid Levels

Participants who identified themselves as non-smokers were asked to fast for at least 6 hours prior to the blood draw. Samples were collected at a General Research Center under low light conditions and protected from light throughout processing [38]. The sera were stored at -70 degree C until analysis by a high-performance liquid chromatography method that was used to measure concentrations of alpha-carotene, beta-carotene, lycopene, lutein, zeaxanthin, and beta-cryptoxanthin, by the laboratory at Genox Corporation, Baltimore, MD.

Theoretical Constructs

Self-efficacy for F&V consumption was measured using a 6-item scale [56]. Participants were asked how confident they were about eating F&V in six different situations and responded on a 5-point scale: (1 not at all confident) and 5 (completely confident). GST constructs were assessed with a goal assessment battery that included five measures that were designed for this study because no scales existed. The battery included (1) identification of current life goals or priorities, (2) evaluation of success in meeting life goals, (3) evaluation of how current life goals or priorities previously selected conflict or facilitate healthy eating, (4) evaluation of life goal pursuit targeting the top life goal or priority that most interferes with eating a healthy diet, and (5) evaluation of dietary goal pursuit, respectively. First, participants were given a list of 15 life common goals (see Table 3 in Multimedia Appendix 2) and asked to identify those they were currently and actively trying to achieve (“goals or activities that you are spending time and effort on at least weekly”). The goals were ranked by the participants on a scale from 1-10 where 1 is not at all important and 10 is extremely important. Next, participants evaluated their success in meeting the goals by responding to the following question for each life goal: “How successful do you feel in meeting this goal right now?” Participants ranked each goal on a scale from 1-5 where 1 is not successful and 5 is extremely successful. Goal conflict and facilitation were measured by asking how the goals affect success in eating a healthy diet (“does the goal make it easier or harder for you to eat a healthy diet?”). The goal was ranked on a 5-point scale where 1 is much easier and 5 is much harder. An example of a question is “On a scale of 1 to 5, does getting more education or another degree make it easier or harder for you to eat a healthy diet?” Last, the construct of goal pursuit was assessed by asking the participant to evaluate the top life goal or priority that most interferes with eating a healthy diet. Participants selected the one priority or goal that interfered the most with eating a healthy diet, and they were asked to respond to a series of statements about pursing that goal (working toward this goal is exciting for me; I receive a lot of encouragement for working on this goal). Responses were on a scale where “1 is not at all true for me” and “5 describes me very well”. Goal pursuit for healthy eating was assessed last. Participants were asked to rate how well each of 29 statements described them: “I want you to think about the goal of eating a healthy diet. Please think about the goal of eating a healthy diet, and tell me how well each of these statements describes you. Please rate on a 1 to 5 scale where 1 is not at all true for you and 5 means it describes you very well.” Some sample items were “Working toward this goal is exciting”, “I try not to let other goals interfere with this goal”, and “I reward myself for working hard on this goal”.

Cost Assessment

Data were collected in order to complete a cost-effectiveness analysis of the maintenance interventions (see Data Analysis section). Direct costs were measured and included the TLC system costs (hardware, software, telephone, and labor). Development costs are excluded. The direct cost of implementing the interventions (TLC) was estimated by tracking the time a research assistant spent on tasks that would occur if the intervention were implemented outside of a research study (ie, labor costs of training personnel and operating and maintaining TLC). All tasks were categorized and tracked on the research assistants' computers. Time spent on tasks that involved training the participant how to use TLC, telephone calls to assist the participant with TLC and computer server were tracked in a computer program. Research assistants logged onto their research operations system at the start of their workday, and any appropriate tasks were tracked by the system. Tasks that were specific to the research study were not tracked, such as assessment phone calls.

Sample Size

Sample size for the RCT was based on 80% power to detect a 20% difference in the percentage maintaining an improvement in F&V consumption at 24 months. This sample size also provides 80% power to detect a small-to-medium effect size for differences in mean F&V consumption based on Cohen's definition [57]. The primary hypothesis was that the treatment groups would maintain their initial gains in F&V consumption better than the control group, and the TLC-GST group would be significantly more successful in achieving this outcome than the TLC-SCT group (ie, GST > SCT > Control). This was tested defining maintenance as sustaining a one-serving increase in F&V that was achieved during the acquisition phase (as this is the eligibility criterion for the RCT). This primary hypothesis was tested using two approaches: a categorical approach and a continuous approach. The categorical hypothesis tests the proportion of the group who maintain, while the continuous tests the differences in F&V serving size. For the categorical hypothesis, the sample size estimation was based on a 20% difference in the proportion of participants meeting the definition of maintenance between groups. If the proportion maintaining gains is 80% in one group, 60% in another, and 40% in a third, there is 80% power of detecting pairwise differences between groups. Expecting 5% attrition at every 6 months from baseline to 24-month time point, a sample size of 405 (n=135 per group) is required at baseline and will yield a final sample of 330 (n=110 per group). This sample size provides 81% power of detecting the difference at 24-month time point between TLC-SCT and Control Groups (60% vs 40%), 87% power of detecting the difference between TLC-GST and TLC-SCT Groups (80% vs 60%), and 99% power of detecting the difference between TLC-GST and Control Groups (80% vs 40%), testing at the two-tailed alpha .05 level. For the continuous hypothesis, group differences are measured by the mean change from baseline to 24-month time point. A sample size of 110 provides 80% power of detecting an effect size of .38 of the standard deviation of the change score in F&V servings.

Results

Data Analysis

Data will be analyzed using SAS, version 9.1 for Windows. Alpha level was set at $P<.05$. Descriptive statistics will be used to characterize the different study subject samples before and after the acquisition study period. Descriptive statistics will examine those individuals who were screened for eligibility and those who qualified and enrolled into the RCT. Sample comparisons between those who were eligible and those who were not will be made using data collected at enrollment (screening data). Comparisons of the different study samples will be performed using Student's *t* test for continuous variables and Pearson's chi-square test for categorical variables. An additional analysis will be performed evaluating the change in F&V servings from initial screening (enrollment) to baseline (ie, the beginning of the maintenance phase) for the sample who was randomized into the RCT. An intention-to-treat approach using the last observation carried forward approach will be used to include those participants who drop out of the study or for whom there are missing data.

The primary hypotheses for the RCT is that the treatment groups will maintain their initial gains (ie, one-serving increase) in F&V consumption better than the control group, and the TLC-GST group will be significantly more successful in achieving this outcome than the TLC-SCT group (ie, GST > SCT > Control) on both a categorical (primary hypothesis) and a continuous measure (secondary hypothesis) of maintenance at all follow-up time points. The primary time points for the study will be baseline (randomization), 6 months (post intervention period), and 24 months (end of the follow-up period). The primary comparisons will be considered from baseline to 6 months, and baseline to 24 months. Group differences will be compared using a continuous variable (F&V consumption) at these time points. We will also compare groups on the percent who maintained a one-serving/day or greater increase in F&V consumption. The categorical measure of maintenance will be analyzed through multiple logistic regression models, and a continuous measure of maintenance will be analyzed through multiple linear regression models. Independent variables in these models will include a set of indicator variables for study group, baseline levels relating to each outcome variable, and potential confounders (variables found to differ between groups at baseline).

Longitudinal models will be used to explore group differences on dietary indicators over time, using data from all study evaluation points. These analyses will use the Generalized Estimating Equations (GEE) approach to accounting for the longitudinal nature of the data by modeling the within-subject correlation and adjusting both regression parameters and standard errors for this correlation. As compared to traditional repeated measures analysis of variance, the GEE approach allows for the inclusion of all available data from subjects with incomplete follow-up in the analysis. For categorical outcome measures, GEE logistic regression models for longitudinal data will be used, while for continuous outcome measures, GEE linear regression models will be used. Independent variables in

these models will include a set of indicator variables for group, a set of indicator variables representing time (with baseline taken as the reference group), and a set of interaction terms modeling differential changes over time for the three study groups. Potential confounders identified in preliminary analyses will also be included in these models. Our primary interest is in the interaction terms, which will test whether the pattern of change in consumption of F&V over time differs by group.

While the primary focus of our analyses of the number of F&V servings is on changes from baseline to 6 to 24 months, our longitudinal models will also allow us to examine maintenance decay in the number of servings from 6 months (at the end of the maintenance intervention) to 24 months. Similarly, our longitudinal models for the categorized maintenance outcome will allow us to examine changes in maintenance from 6 to 24 months as well.

Secondary analyses will examine the influence of psychosocial variables. For TLC-SCT, we hypothesize that the outcomes are mediated by self-efficacy. For TLC-GST, we hypothesize that outcomes are at least partially explained by changes in goal system variables such as levels of inter-goal facilitation, inter-goal substitution, and inter-goal conflict at all major follow-up time points.

These hypotheses will be tested with path analysis. Potential mediational pathways of the effect between the randomized groups and F&V intake for the theoretical construct variables will be examined. Path models will be constructed to test the direct and indirect associations indicated by our research model. Using 6 and 12 month data as indicators of processes of maintenance in both the mid-term (at the end of the maintenance intervention) and in the longer term (6+ months post intervention), path analyses, both separate and combined, will be conducted to examine mediators of maintenance variables as influenced by group assignment. To examine the TLC-SCT change model, paths will be modeled from an intervention variable to self-efficacy to F&V intake. Direct and indirect effects of the intervention will be estimated through standardized path coefficients. For the TLC-GST, goal system variables will be examined in analogous path models.

Cost Analysis

Cost analyses are planned for the study conditional upon demonstrating that the maintenance interventions are effective in altering and sustaining improvements in diet. The analysis will be based on the recommendations of the US Public Health Service Panel on Cost-Effectiveness Analysis [58]. An incremental cost-effectiveness ratio on the acquisition phase will be computed. The incremental cost-effectiveness of the two maintenance intervention conditions will be compared relative to the acquisition intervention to assess the resource use associated with incremental sustained improvements in health.

Discussion

Principal Considerations

There are a number of important issues to consider about this study. The overarching goal of this study is to better understand how to help individuals maintain a newly acquired health behavior. The challenge was first to identify a population that is engaging in unhealthful behaviors and that the behavior is amenable to change. F&V was selected as the principal target for the intervention study for multiple reasons. First, the majority of the US population is not consuming enough F&V, providing an opportunity to recruit a large enough sample within a reasonable time frame. Second, we had to identify an evidence-based, off-the-shelf intervention that would likely produce a change in F&V in order to study whether a newly acquired behavior can be sustained with interventions specific to the maintenance of that newly acquired behavior. While the TLC-EAT intervention was used for the acquisition phase of the study, we had to consider how much time it would take to achieve a primary intervention effect using TLC-EAT to make a meaningful positive dietary behavior change to qualify sufficient numbers of participants for the maintenance intervention RCT, and what should be considered a positive response in this RCT. One option was to offer TLC-EAT acquisition intervention for as long as it took the participant to show a positive improvement. This was not feasible given the time limitations of the study but fortunately was not a factor in the study design because the TLC-EAT acquisition intervention was known to achieve a positive effect within 3 months based on a previous study [31]. We also decided to define a positive intervention effect in the acquisition phase as an increase in F&V consumption by one or more servings per day. We did consider using a threshold criterion of a successful acquisition intervention effect of 5 servings a day but did not want to exclude individuals who might begin the acquisition phase at very low levels of F&V consumption and have a substantial acquisition intervention effect but still not meet the threshold of 5 servings of F&V/day. Moreover, including individuals who increase at least one F&V serving a day is in line with the usual level of change achieved in successful dietary change programs [6]. In addition, there is no evidence in the literature that increasing F&V consumption from 0-1 serving/day confers different risk reduction than going from 4-5 servings/day. Moreover, it is not known if it is easier to maintain a one-serving increase from 4-5 servings per day compared to a change from 0-1 servings per day, or whether a larger increase in F&V consumption (for example 2 or more servings/day) is more difficult to maintain than a smaller increase during behavior change acquisition.

A major consideration for our study was how to define maintenance. There is not a well-accepted definition of maintenance of a behavior change. It could be defined as a criterion outcome, which means that the individual consumes at least the same number of servings of F&V at each outcome measurement point as he or she consumed at the end of the acquisition phase. Another approach is to compare the three groups at each time point using the proportion of participants in each study group who reach criterion at that time (eg, consume at least the same number of servings of F&V at the end of acquisition to the end of the maintenance assessment). In doing so, we will consider the standard error of the measurement of F&V intake so that maintenance of F&V consumption will be defined as at least the same intake the person achieved at the end of the TLC-EAT acquisition period

minus the standard error. Another approach is to compare changes in intake of F&V between randomization and each outcome measurement point. These "change scores" can be compared across the three study groups. The advantage of this approach over the criterion outcome approach is that it considers both the degree of preservation of intervention effect and possible increases in intervention effect by the maintenance intervention. Recent research suggests beneficial health effects as F&V consumption increases rather than only among those who meet a certain threshold (ie, 5 servings/day) [2]. The advantage of using a "change score" rather than a "threshold score" is that it may be the most relevant public health criterion. However, it is a less intuitive measure of maintenance than the criterion measure. Thus, both approaches will be examined.

The last consideration concerns the diet content of the basic TLC-EAT intervention. TLC-EAT has shown to improve F&V in two previous interventions yet the topics covered in the intervention calls include other food groups (diary, protein, grains). The approach taken in TLC-EAT relies heavily on the strategy of substitution of "healthy" for "unhealthy" foods; in doing so, it promotes greater consumption of some food groups, like F&V, possibly at the expense of others, like red and processed meats, and recognizes the public health benefits of targeting multiple dietary risk factors and the practical realities of intervening on diet. Thus, the diet topics in all of the interventions were written using this approach.

Strengths

While few studies have evaluated maintenance-specific interventions for dietary change, fewer maintenance-specific interventions have been studied in which outcomes exceed 12 months, and even fewer have compared theory-guided interventions. The major innovation of this project is the design, creation, and testing of a maintenance-specific intervention (TLC-GST) and its comparison with an intervention based on the most commonly used behavioral theory, SCT. GST is a "maintenance-focused" theory in that it posits that many worthy and attainable goals fall to the wayside because other endeavors, for any number of reasons, take resources away from their pursuit. If it is competition for internal and external resources that triggers a significant proportion of relapse, then an intervention that successfully advocates for the continued allocation of resources to dietary behavior, as well as assists users in more efficiently managing the resources they have across their goals, should make a difference. It is also possible that maintenance of goal behaviors that do not have immediate and perceivable rewards, especially true of long-term risk reduction, is especially appropriate for an intervention that helps balance demands from goals that have more short-term and palpable rewards.

Few studies have compared theories, mostly because it is a challenging task that requires that at least two interventions be available that are the same or very similar with each of them based on a different behavioral theory. There are very few pairs of behavioral interventions that have functional comparability while being driven by distinctly different theories. Practically speaking, a researcher has to design the two interventions being studied, and this is time-consuming and expensive. Nonetheless, this is exactly what we did in creating two theoretically guided interventions for maintenance of dietary behavior change. Using SCT to map out a maintenance intervention was doable because it is one of the most widely used theories for changing and maintaining health behaviors. The relapse prevention model [53], which is informed by SCT, provided a practical framework for implementing a SCT-based behavior maintenance intervention by explicitly identifying which intervention strategies to include (eg, goal setting, planning ahead) to maintain a behavior change. The resulting TLC-Maintenance SCT intervention has good fidelity to SCT. In contrast, and to our knowledge, GST has never been applied to health behavior change, whereas it has a strong theoretical and experimental underpinning for application to health behavior and its change. No study has attempted to translate and apply GST for practical dietary interventions delivered to individuals over the telephone.

Another strength of the study design in our RCT is our ability to control for fidelity in delivering the interventions. The acquisition intervention, TLC-EAT, was delivered to all enrolled participants in the same way because it was delivered using an automated computer system. TLC-EAT is designed to provide a consistent intervention of all users that is tailored to the participant. TLC-Maintenance SCT also provides a consistent maintenance intervention for all of its users. Although a computer-controlled health behavior change intervention may or may not be as tailored as a human counselor intervention, the intervention content is explicit and discoverable and has complete fidelity. If differences are found between the two health behavior change maintenance interventions, they will be due to the underlying content of the interventions and not due to differences in provider training, provider skill level, or lack of adherence to protocol [59].

Conclusions

This study used a novel study design to initiate and then promote the maintenance of dietary behavior change through the use of an evidence-based fully automated telephony intervention. After the first 6 months (the acquisition phase), we will examine whether two telephony interventions built using different underlying behavioral theories, were more successful than an assessment-only control group in helping participants maintain their newly acquired health behavior change.

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Conflicts of Interest

None declared.

Multimedia Appendix 1

CONSORT-EHEALTH checklist V1.6.2 [59].

[PDF File (Adobe PDF File), 989KB - resprot_v3i4e62_app1.pdf]

Multimedia Appendix 2

Supplementary tables.

[PDF File (Adobe PDF File), 72KB - resprot_v3i4e62_app2.pdf]

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Abbreviations

F&V: fruits and vegetables
GST: Goal Systems Theory
IVR: interactive voice response
NCI FVS: National Cancer Institute Fruit and Vegetable Screener
RCT: randomized controlled trial
RPM: relapse prevention model
SCT: Social Cognitive Theory
SRC: University of Rhode Island's Survey Research Center
TLC: telephone-linked care
TLC-EAT: telephone-linked care delivered dietary acquisition intervention
TLC-GST: telephone-linked care delivered dietary maintenance intervention based on Goal Systems Theory
TLC-SCT: telephone-linked care delivered dietary maintenance intervention based on Social Cognitive Theory

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Protocol

Pain Management in Cancer Patients Using a Mobile App: Study Design of a Randomized Controlled Trial

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Abstract

Background: Despite the availability of effective medications and clinical guidelines for pain management, pain control is suboptimal in a sizeable proportion of patients with cancer pain. The National Comprehensive Cancer Network guidelines recommend a comprehensive and multimodal approach for management of cancer pain. We developed a mobile phone application, ePAL, based on clinical guidelines to empower patients for cancer pain management by prompting regular pain assessments and coaching for self-management.

Objective: The objective of this study is to evaluate the effect of a multidimensional mobile phone-based pain management application, ePAL, on controlling cancer pain and improving quality of life in patients with cancer pain being treated at an academic palliative care clinic.

Methods: The study will be implemented as a 2-arm randomized controlled trial with 110 adult patients with CP who own a mobile phone over a follow-up period of two months. Participants will be randomized to either the intervention group receiving ePAL and usual care or to a control group receiving only usual care. The brief pain inventory will be used to assess our primary outcome which is pain intensity. We will also evaluate the effect of the intervention on secondary outcomes which include the effect of the intervention on hospital utilization for pain crisis, quality of life, adherence to analgesic medications, barriers to pain control, anxiety and patient engagement. Instruments that will be used in evaluating secondary outcomes include the Brief Pain Inventory, Morisky Medication Adherence Scale, Barriers Questionnaire-II, Functional Assessment of Cancer Therapy–General, Edmonton Symptom Assessment System, Generalized Anxiety Disorder 7-item scale, and the Functional Assessment of Chronic Illness Therapy-Fatigue. The intention-to-treat approach will be used to evaluate outcomes. Our primary outcome, pain intensity, measured longitudinally over eight weeks, will be assessed by mixed model repeated analysis. Effect sizes will be calculated as mean group differences with standard deviations.

Results: The study is still in progress. We hope to have results by the end of 2015.

Conclusions: The multidimensional approach to pain management implemented on a mobile phone application could lead to significant improvements in patient outcomes.

Trial Registration: ClinicalTrials.gov NCT02069743; https://clinicaltrials.gov/ct2/show/NCT02069743 (Archived by WebCite at http://www.webcitation.org/6Qb65XGGA).

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KEYWORDS

cancer pain; mobile application; randomized controlled trial; self care; palliative care; mHealth

Introduction

Background

In addition to the inherent pathophysiological effects associated with any cancer, pain is one of the most feared consequences of cancer [1,2]. Cancer Pain (CP) may be present at any stage of the disease process. It may be the first complaint patients experience before diagnosis and may even be present long after treatment. Pain may be due to the direct effect of the cancer, diagnostic procedures or treatment. It is associated with distressing psychosocial discomforts and adversely impacts the patient's quality of life [1]. The prevalence of CP varies depending on the type of cancer and stage of the disease; it ranges from 24%-60% in patients receiving treatment, 62%-86% in patients with advanced cancer, and 33% in patients post curative treatment [2-5]. Results from a meta-analysis showed that the prevalence of CP was 70%, 59%, 55% 54%, 52%, 60%, and 50% in head and neck, gastrointestinal, lung/bronchus, breast, urogenital, gynecological, and all cancer types respectively [6].

Despite the availability of guidelines from organizations such as the World Health Organization (WHO), at least 14% of patients with cancer pain have inadequate analgesia [7,8]. Barriers to the adequate management of CP may be physician or patient-related. Guidelines recommend that patients be screened (with standardized assessment scales) for CP at any encounter with their providers, but physicians generally under-assess pain, given their busy clinic schedules. Additionally, while some physicians have misconceptions about drug tolerance, addiction, routes of administration, and exaggerated fear of adverse effects that accounts for the sub-optimal management of CP [9]. Poor patient-physician communication is another reason for failure to accurately establish the intensity of the patient's pain. In the United States, this is especially worse in ethnic minorities and contributes to further widening the health disparity gap [10,11]. Many patients want to be "good" patients and do not want to "trouble" their physicians. As a result they may have misconceptions about "pain as an unavoidable consequence of cancer", fear of addiction, issues of medication compliance, and side effects from their pain medication [9]. These provider and patient-based barriers can culminate in a burden on the patient-provider relationship and result in inadequate treatment of cancer pain.

Cancer pain is multidimensional in nature and requires intensive self-management. It is important for affected patients to be coached, educated, and empowered to manage the pain they experience. Unfortunately, randomized clinical trials have been scarce on this subject [1]. The standard care in pain management is based on the analgesic ladder recommended by WHO from which most other guidelines have evolved. Although the guidelines have shown to be effective, evidence suggests that they are poorly implemented in practice [12]. Gaps in knowledge about pain, clinical applications of pain, and a perpetual disconnect between patient and provider regarding pain, are all impeding progress in developing best practices for CP management. The National Comprehensive Cancer Network (NCCN) guidelines recommend a comprehensive and multimodal approach for management of CP [3]. Pain can be controlled if recommended algorithms are systematically applied and tailored to meet patients' needs [3].

Traditionally, management modalities are administered during visits to a clinician be it an oncologist, nurse or social worker. However, we developed a self-management mobile phone application, ePAL that patients can use in between visits to manage CP. The tool was developed based on clinical guidelines to empower patients for self-management by facilitating patient education, regular pain assessments, and timely feedback, medication changes and improving patient-physician connections. Our strategy is focused on supporting patients by regularly nudging them to assess their level of pain control and coaching them about CP management through regular messaging.

Our goal is to help patients with cancer pain gain better control of their symptoms and improve their overall quality of life. Therefore, we hypothesize that patients randomized to receive ePAL will have better pain outcomes, measured by pain intensity, and improved quality of life compared with patients receiving usual care.

Specific Aims

Primary Aim

Our primary aim is to evaluate the effect of ePAL on pain control in patients with cancer pain.

Secondary Aims

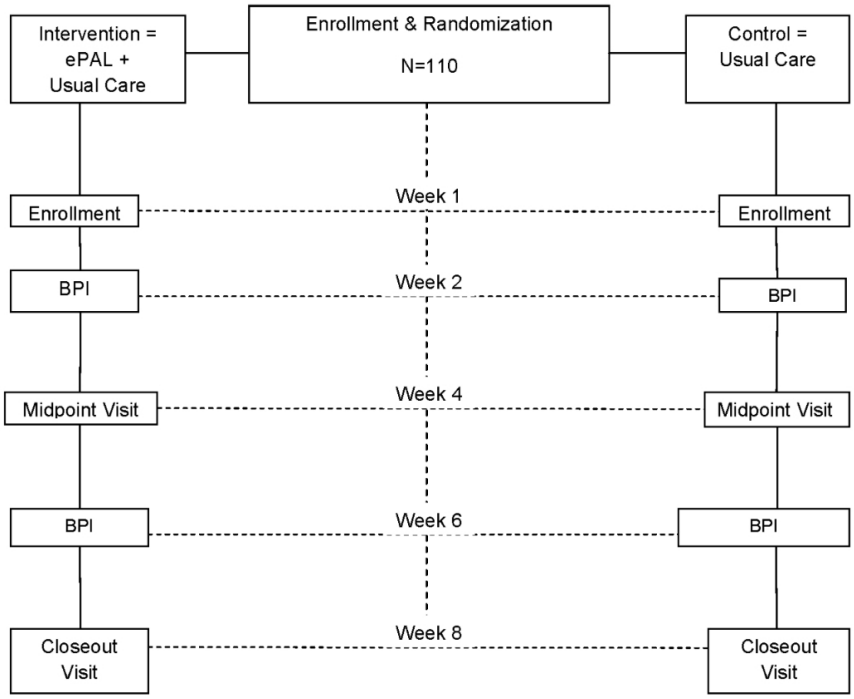
Secondary outcomes to be assessed include evaluating the effects of ePAL on: (1) quality of life; (2) hospital utilization for pain crises; (3) adherence to analgesic medications; (4) daily opioid consumption; (5) levels of anxiety; and to evaluate (6) participants' pattern of engagement with ePAL; and (7) perceived barriers to cancer pain management.

Methods

Trial Design

This study will be implemented as a 2-arm randomized controlled trial (RCT; NCT02069743) with repeated assessments at baseline, midpoint, and at the end of the study over an eight weeks follow-up period. Figure 1 shows the research design.

Figure 1. Schematic summary of the trial design. BPI: Pain intensity assessments with the Brief Pain Inventory.



Participant Inclusion/Exclusion Criteria

Patients must meet all eligibility requirements to be enrolled into the study. Eligible patients are ambulatory, patients aged 18 years or older with diagnoses of a solid-organ cancer and moderate to severe levels (at least 4/10 on the numerical rating scale [NRS]) of pain at enrollment. They must also have a mobile phone and should be able to read and speak English. Ineligible patients will be defined as those who (1) have a life expectancy less than two months as determined by the clinical team; (2) have any significant medical or psychiatric comorbidities (other than depression or anxiety) or cognitive impediments that would prevent them from being able to utilize the program; (3) have a known history of substance abuse; and (4) participating in any other investigational therapies or other study protocols that may have an impact on pain intensity or quality of life which are the main outcomes of this study.

Recruitment Procedure

All patients, regardless of ethnicity, with upcoming appointments at the Massachusetts General Hospital (MGH) Palliative Care Center are considered potential candidates. Therefore, potential subjects for this study will be drawn from adult cancer patients presenting with pain at the MGH Palliative Care Center. They will undergo screening to ensure eligibility and formal enrollment by signing the informed consent form. This is done before any study procedure is carried out. Participating subjects will complete all enrollment surveys and randomization procedures. All participants will be instructed to continue to receive medical care from physicians as usual. Subjects randomized to the intervention arm download the ePAL mobile application and will be taught how to use the functions of the application.

Intervention

Framework for Intervention

Overview

The ePAL mobile application was designed to augment existing pain management protocols at the MGH Palliative Care Center and to promote self-efficacy for pain management in patients with cancer pain. Therefore, we use a comprehensive and multimodal approach as recommended by the NCCN guidelines for treatment of adults in addition to addressing common challenges in managing CP, as identified in oncologic clinical practice and in the medical literature [3]. The recommended targeted dimensions for CP management include providing support, encouraging subjects to report CP, teaching coping skills, and building self-efficacy. This app is based on the premise that providing a platform where patients can easily evaluate pain control and find support to manage their pain will overall improve pain management and quality of life. Therefore, ePAL (1) provides a medium for on-demand pain assessments using a NRS (ie, patients can use the app to assess the level of their pain control at any time when they experience exacerbation of symptoms or breakthrough pain); (2) coaches patients using bite-sized educational and supportive messages; (3) facilitates patient-provider communication; (4) helps patients track their symptoms, and (5) requests prescription refills. Furthermore, the intervention is tailored to the individual patient's need which is determined by the ratings of pain intensity on the NRS, self-reported symptoms and self-reported barriers to pain management. These factors were taken into account in the design of the app and aided in determining the type of educational coaching messages that a particular subject receives. We further elaborate on our approach below.

Increasing Opportunities for Pain Control

Pain control is routinely assessed when a patient complains about it during a scheduled visit to the doctor's office (every 2-6 weeks, depending on patient's clinical condition) or during unscheduled patient-initiated visits for uncontrolled pain. ePAL on the other hand, can be easily administered outside of hospital settings. The app prompts participants to assess the level of their pain control over the past 24 hours three times per week. As a result, it provides patients with a medium to regularly evaluate pain control and opportunities for management. Participants with breakthrough pain or other CP-related complaints are able to perform ad hoc pain assessments and management at any time. The app recommends management strategies based on responses to the pain assessment and other pain management barriers. Treatment recommendations are not prescriptive but guide patients on what to do to help them manage their pain. Patients with newly occurring, persistent, or severe pain will be connected directly to the MGH Palliative Care Center for further evaluation and treatment. This is enabled by the automatic call function on the app that links directly to the MGH Palliative Care Center. However, patients have the option to decline speaking with the care team about the trigger symptom. For patients who need further evaluation outside of the regular pain clinic hours, they will be automatically connected with the Palliative Care on-call physician. In addition, participants can pursue medical assistance through the usual emergency phone and paging protocols at the Mass General Palliative Care Center and Cancer Center.

Coaching

Our hope is that by the time patients graduate from the program, at the end of the 2-month study, they will be better equipped to manage their CP in between their clinic visits or appropriately report uncontrolled pain. Contents from the educational library of ePAL will be used to coach participants on pharmacologic and nonpharmacological interventions to reduce CP. Coaching will also address the various misconceptions associated with CP management [12,13]. Patients are coached through small, bite-sized push notification with daily "tips" for managing cancer, cancer pain, and other side effects. These messages will help keep patients connected to their care providers even when they are not immediately due for another clinic visit, and facilitate improved communication between patients and providers. Additionally, coaching will also address medication adherence which will be encouraged by emphasizing the need for "by the clock" administration, addressing misconceptions about tolerance and addiction, managing side effects of analgesic therapy, and providing general education about cancer and CP [11-14].

Key Features of ePAL

Pain Assessments and Feedback

This is one of the key features of the app. Patients are nudged to assess level of their pain control three times per week. However, they are also able to perform ad hoc pain assessments apart from these scheduled prompts. Patients input the level of their pain control on a sliding NRS on the homepage of the ePAL. After patients rate their pain levels, the app will assess whether it is a new or recurring pain, the acceptability or tolerability of the pain, barriers to pain control and also rule out red flag symptoms (eg, persistent vomiting, no bowel movement for >3 days) that may require further evaluation by the care provider. Based on the identified barriers to adequate pain control, the app then suggests management steps and coaches them by addressing the identified barriers.

Multimedia Library

Patients often report a need for easily accessible and understandable information. For the ePAL mobile app, we developed a comprehensive library with information some of which were created by the investigators and others chosen from reliable sources. The library contains information on a wide array of topics relating to cancer, cancer pain, pain medication, side effects of pain medications, and other related symptoms, barriers to pain management, etc. The library is broken down into topic pages to facilitate users learning at their own pace instead of being overwhelmed by too much information. The app also includes videos (featuring physicians and nurse practitioners from the MGH Palliative Care Center explaining important topics in CP management) and audio files on relaxation and meditation techniques for pain management.

Notebook

The app offers a diary, directly accessible from the homepage and other relevant locations, for patients to log their experiences dealing with their symptoms and questions they have about their management protocols. This was incorporated into the app based on exploratory needs assessment where some patients reported having lots of questions for their providers which they forget to ask at the clinic visit and leave without hearing the answers. Patients can generate a report of all their notes and bring them up for discussion during the in-person visit. We hope the notes will help the patients better organize their discussions with their care providers during the clinic visits.

Tracking

The app tracks pain scores so that patients can monitor their progress and also share their trends with their care provider and support system. There are two available views: a sliding weekly scale and a monthly calendar view. Pain scores are displayed in green, yellow, and red which correspond to mild, moderate, and severe pain respectively. From here, users can also directly access notes associated with each day.

Medication Refill Request

This is an additional feature to bolster user engagement with the app and also to facilitate continuous pain management without the interruption of unfilled prescriptions. This is based on findings from exploratory needs assessment at the MGH Palliative Care Center where patients reported that calling the hospital for refills, pain consultations, appointment scheduling, etc could be stressful and time-consuming. Patients can request refills in one simple tap on the app.

Screenshots of the ePAL mobile intervention are shown in Figures 2 and 3. Figure 2 shows the welcome screen of the application and a sample of educational messages (a), the resulting feedback from a pain score of >8 (b, c), and the symptom tracking function (d). In Figure 3, the additional features of the intervention are shown. These include the ability

to order prescription refills and make notes for tracking and or with specific questions for the physician.

Figure 2. ePAL Mobile Application. a-d shows the numerical rating scale and pain tracking feature: a). Welcome page with educational tip; b & c). NRS and resulting message if pain score >8; d). Trend of pain scores, with notes.

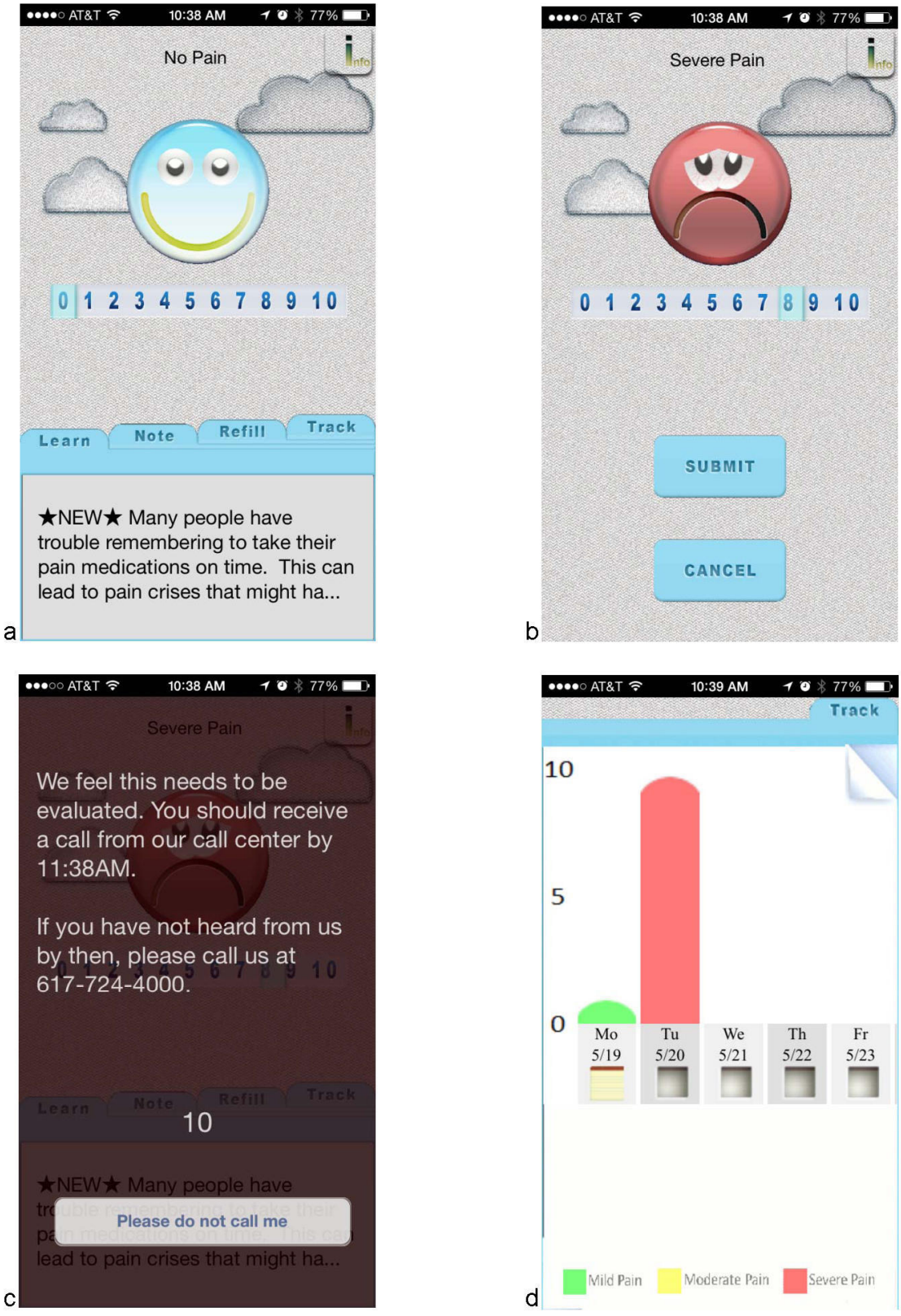
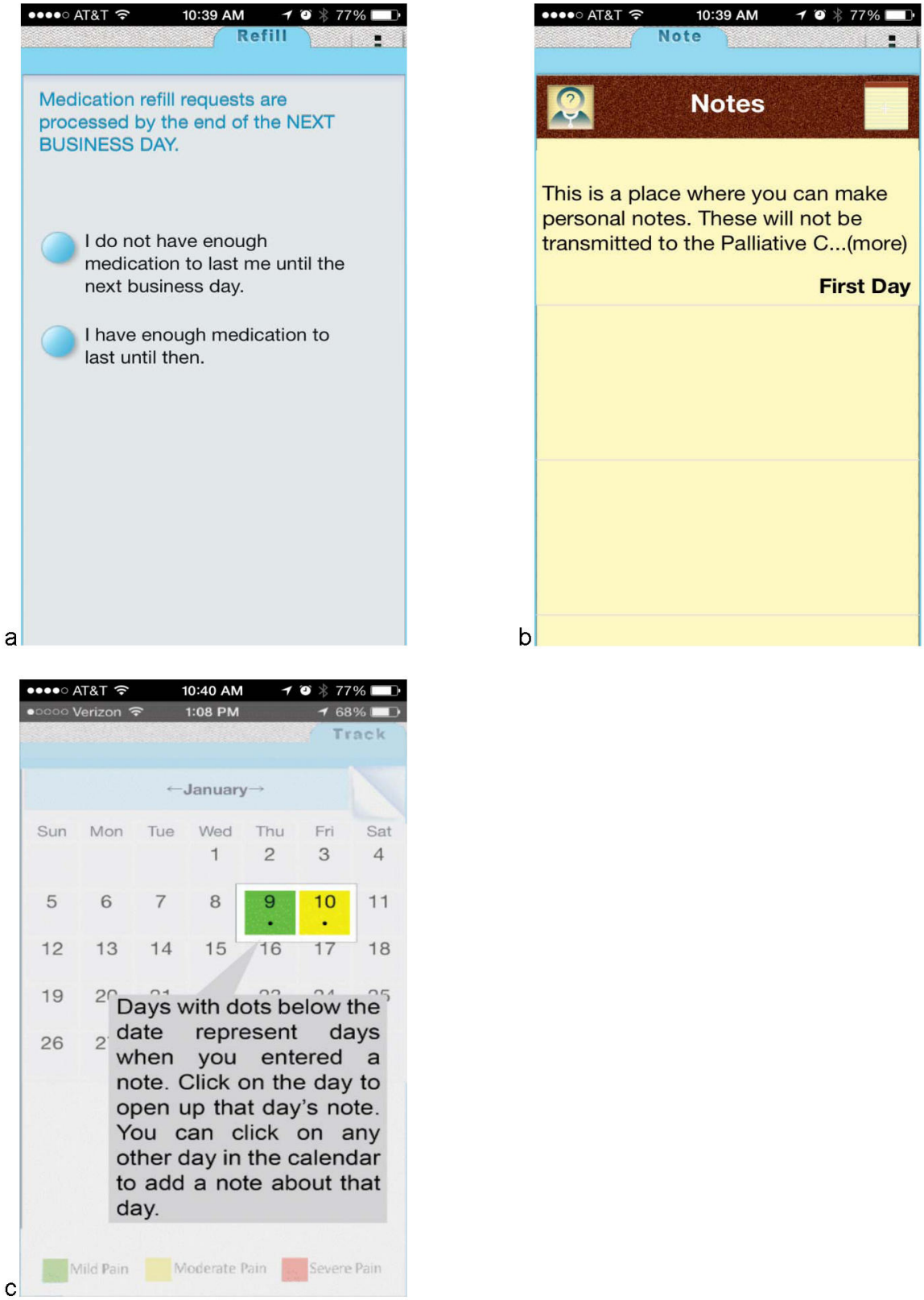


Figure 3. ePAL Mobile Application Features: a). Prescription refill; b). Notes feature; c). Calendar showing days with notes.



Intervention Group

Participants will continue to receive medical care from their oncologist, palliative care providers, primary care physicians and other care teams as usual. In addition to usual medical care, they will be able to use ePAL to manage their pain. During the study, intervention subjects will receive push notifications on their phones to assess their pain control over the past 24 hours every morning on Mondays, Wednesdays, and Fridays throughout the study period. The app will guide them through

management steps based on their responses. Additionally, one coaching message will be sent to subjects once every day. Subjects will also be encouraged to use the app to manage their pain on-demand, and also to use other features of the app, including pain tracking, prescription refill requests, and the multimedia education library.

Control Group

Participants in the control group will continue to receive medical care from their care teams as usual. However, they will not be able to use the app during the study period.

Outcome Measures

Data Collection Materials

There is one primary outcome and several secondary outcomes that will be assessed in this trial. This study uses several validated instruments for data collection: (1) the Brief Pain Inventory (BPI) assesses participants' level of cancer pain and interference with daily function [15]; (2) the Morisky Medication Adherence Scale (MMAS-8) is an 8-item self-reported questionnaire used to measure adherence to medication [16]; (3) the Barriers Questionnaire-II (BQ-II) is used to capture subjects' beliefs that may impact optimal pain control [17]; (4) the Patient-Health Questionnaire PHQ-8 is used to screen for depression [18]; (5) the Functional Assessment of Cancer Therapy-General (FACT-G) is used to measure health-related quality of life [19]; (6) Edmonton Symptom Assessment System (ESAS-r) is used to assess symptoms commonly found in palliative care patients [20]; (7) the generalized anxiety disorder 7-item scale (GAD-7) is an easy to use instrument to assess levels of anxiety [21]; (8) the Functional Assessment of Chronic Illness Therapy-Fatigue (FACIT-F) assesses levels of fatigue in patients with chronic illnesses [22].

Primary Outcome

Primary outcome measurement is pain intensity measured as a continuous outcome. It will be measured quantitatively with the BPI that assesses worst, least, average, and current pain intensity over the past week. The self-administered paper version will be used during in-person visits at baseline, midpoint, and at closeout; while the Interactive Voice Response (IVR) version will be used at weeks 2, 4, and 6.To minimize loss of data, any subject who is not able to complete the BPI at weeks 2 and 6 via IVR will be sent REDCap links to complete the surveys online. REDCap is a secure, Web-based application for data collection customizable for individual research studies. It is free and complies with all Health Insurance Portability and Accountability Act regulations (HIPAA). It was developed by a multi-institutional consortium initiated at Vanderbilt University.

Secondary Outcomes

All secondary outcomes will be assessed at midpoint and at the end of the study. Quality of life will be assessed with the FACT-G and ESAS-r. Hospital utilizations for pain crisis including emergency department or urgent care visits and unplanned clinic visits for pain will be assessed from the Electronic Medical Records. In addition, adherence to analgesic medications will be measured by the MMAS-8; opioid consumption will be measured quantitatively as oral morphine equivalent daily dose; patient-related barriers to pain will be measured quantitatively by the BQ-II; anxiety will be measured by the GAD-7; and pattern of patient engagement with ePAL will be assessed quantitatively by subject's interaction with the system measured by the number of their responses to interactive messages and the use of other functions such as the notes function on the app.

Data Collection

Data will be collected at various time points (baseline, weeks 2, 4, 6 and 8) as described in Table 1. Data collection for weeks 2 and 6 will be via the IVR system while baseline, midpoint, and closeout visits will be conducted in-person. However, if for unavoidable reasons, in-person visits are not possible, survey questionnaires will be sent via REDCap. Outcome data will be stored in computer network files that are accessible only to IRB-approved study staff. Outcome data will be linked to each subject by a unique number, which will not have any identifying information. All data collected will be analyzed at the end of the study period.

Table 1. Data collection schedule: the table depicts the schedule for data collection.

Intervention/Control group data collection	At entry	2 weeks	4 weeks (+/- 5 days)	6 weeks	8 weeks Closeout (+/- 5days)
Enrollment questionnaire	X				
PHQ-8	X				
BPI	X	X	X	X	X
MMAS	X		X		X
BQ- II	X		X		X
FACT-G	X		X		X
ESAS-r	X		X		X
GAD-7	X		X		X
FACIT-F	X		X		X
Opioid consumption	X		X		X
Hospital utilizations					X

Statistical Analysis Plan

Sample Size Estimation

A sample size of 88 subjects, 44 in each arm, is sufficient to detect a clinically important difference of 1.5 between the control and intervention arms in pain intensity scores assuming equal standard deviation of 2.5, using a two-tailed *t* test of difference between means, with 80% power and a 2-sided alpha of .05. Considering a dropout rate of 20%, the sample size required is 110 (55 per group). Fifty-five participants will be randomly assigned to the intervention and the remaining 55 participants will be assigned to the standard practice of care at the MGH Palliative Care Center (usual care group). Participants will be followed up for a total of 8 weeks.

Randomization

A computer program will be used to randomize subjects into the intervention or control arm in a ratio of 1:1 using random permutated blocks to optimize balance in each treatment at any given point in time during the study. Treatment assignment will be concealed in sealed envelopes prepared by a third party not directly involved with the study. Treatment assignment will be revealed after the consent form has been signed by the study participant. Due to the nature of this study, we will not be able to blind subjects to their treatment assignments. However, treatment assignments will be concealed from the investigators and the data analyst.

Statistical Analysis

Data analysis will be done with Data Analysis and Statistical Software: STATA, version 13 with an alpha of .05 set a priori for all analyses. All participants will be followed up for eight weeks and we will summarize their baseline demographic and technology use characteristics by study arm. Continuous variables will be compared between control and interventions using the *t* test and categorical variables will be compared using chi-square test. All analyses will be based on intention-to-treat in all randomized participants. Our primary outcome, pain intensity, measured longitudinally over eight weeks, will be assessed by mixed model analysis of variance with treatment assignment as between-group factor and time as within-subject factor. Effect sizes will be calculated as mean group differences with standard deviations. A similar approach will be used to analyze continuous secondary outcomes while categorical outcomes will be analyzed by chi-square test.

Ethics and Informed Consent

Procedures of our methods have been reviewed and approved by the Dana-Farber/Harvard Cancer Center (DF/HCC) Institutional Review Board (IRB) and the study is registered at [23]. The app is secure and complies with all HIPAA requirements. Subjects will require a secure pass code to be able to access the app. All mobile phone numbers are stored in a secure shared drive available only to IRB-approved study staff. However, if any data breach or adverse effect occurs, the investigator will ensure that they are well-documented and reported according to the IRB's requirements, regardless of causality.

Potential candidates with upcoming appointments at the Palliative Care Center will receive a letter informing them about the study. The study consent will also be sent along so that interested candidates can review and ask relevant questions during the enrollment visit. During the study visit, subjects will be given sufficient time to review the consent form and will be encouraged to ask questions. A research assistant will go through the consent form with subjects to ensure they comprehend details of the study. Thereafter, subjects will sign two copies of the consent form–one for them and the other for the study team. Patients who want more time to consider participating in the study will be sent home with an unsigned copy of the consent form and encouraged to think about it at their convenience. Should these patients choose to participate, they will be rescheduled for another enrollment visit. After the informed consent process, the research assistant will then perform all other enrollment procedures.

Discussion

Principal Findings

Our expectation is that enabling patients to assess pain control on-demand and empowering them for self-management will lead to improvements in pain and quality of life outcomes. The ePAL mobile application enables patients to assess pain levels, track progression, manage cancer pain and associated symptoms outside hospital settings. Additionally, the app addresses patients' beliefs that may hinder optimal pain control and also provides an easy way to connect with care providers for timely medication refills. When patients are educated and better understand their cancer and the pain that may be associated with it, improved skills on CP management can be adopted [12,14]. In addition to the capability to generate weekly symptom reports that could be shared with providers, the app also has a notebook function that allows patients to log their symptoms and other relevant questions to ask their care providers at their clinic visits. We hope these functions will facilitate improved patient-provider communication.

Cancer pain, when not managed timely or adequately, may progress to pain crisis necessitating immediate intervention. Pain crisis refers to an episode of severe, uncontrolled pain that causes patients and their caregivers significant distress [9,14]. Epidemiological data on incidence of pain crises in cancer management is scarce but some centers report that pain crises account for about 20-25% of all palliative care referrals [14]. Our intervention has the capacity to identify early on, cancer pain patients that are at risk of experiencing pain crises and eventual hospitalization.

A special concern is that the frequent prompts to assess pain control may cause a heightened awareness of symptoms and increase anxiety in some participants. While this is plausible, we hope that the application will increase self-efficacy for self-management. Therefore, the GAD-7 will be useful in establishing the effect of our approach on anxiety. A study by Kroenke and colleagues showed that a telecare management of pain and depression in patients with cancer actually had a positive effect on anxiety levels and was also associated with improvements in pain control and other quality of life outcomes

in intervention participants [24]. Another similar concern is that the frequent pain assessments may also increase opioid use. Rustoen et al's 6-week trial of PRO-SELF, a self-care intervention for cancer pain management that involved daily logging of pain intensity and analgesic intake, revealed that opioid intake increased significantly in all patients in the study [25]. They also reported that study participants took about 40% higher doses of opioids than reported in previous studies although they speculated that this could be due to the fact that only patients with bone metastases were included in the study [25]. It remains to be determined how ePAL will impact opioid consumption. However, this present study does not require daily logging of pain intensity but suggests pain assessments three times per week.

Limitations

Given that the sample population is drawn from patients attending the palliative care clinic of a large academic center, majority of these patients may have advanced cancers and complicated diseases. As a result, there is a possibility that some patients may not be able to complete the study or attend final study visits. To limit loss to follow-up and missing data, we limit the participation to patients with life expectancy >2 months. Also, we will measure endpoints at multiple times and use REDCap for those unable to attend the final study visit. Another potential limitation is concerning the external validity of the study: This is because findings from this study may be generalizable to patients in academic medical centers. However, if the intervention proves to be effective in these settings, we believe that it will also be effective in patients receiving care in nonacademic medical centers because the app is patient-centered and easily adaptable to other settings.

Conclusions

This is one of the first clinical trials evaluating the impact of a multidimensional evidence-based approach to pain management via a mobile phone application. Our hope is that findings from this study could lead to larger, multicenter trials to demonstrate more robust evidence. We also hope that similar evidence-based approaches can be developed for other acute-on-chronic conditions to empower patients for self-care, reduce cost, and improve overall patient outcomes.

Acknowledgments

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Conflicts of Interest

The authors of this article designed ePAL but are not responsible for the day-to-day running of the trial.

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Abbreviations

CP: cancer pain
HIPAA: Health Insurance Portability and Accountability Act regulations
IRB: Institutional Review Board
IVR: Interactive Voice Response
MGH: Massachusetts General Hospital
NCCN: National Comprehensive Cancer Network
NRS: numerical rating scale
RCT: randomized controlled trial
WHO: World Health Organization

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Protocol

A Telehealth Intervention Using Nintendo Wii Fit Balance Boards and iPads to Improve Walking in Older Adults With Lower Limb Amputation (Wii.n.Walk): Study Protocol for a Randomized Controlled Trial

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Abstract

Background: The number of older adults living with lower limb amputation (LLA) who require rehabilitation for improving their walking capacity and mobility is growing. Existing rehabilitation practices frequently fail to meet this demand. Nintendo Wii Fit may be a valuable tool to enable rehabilitation interventions. Based on pilot studies, we have developed “Wii.n.Walk”, an in-home telehealth Wii Fit intervention targeted to improve walking capacity in older adults with LLA.

Objective: The objective of this study is to determine whether the Wii.n.Walk intervention enhances walking capacity compared to an attention control group.

Methods: This project is a multi-site (Vancouver BC, London ON), parallel, evaluator-blind randomized controlled trial. Participants include community-dwelling older adults over the age of 50 years with unilateral transtibial or transfemoral amputation. Participants will be stratified by site and block randomized in triplets to either the Wii.n.Walk intervention or an attention control group employing the Wii Big Brain cognitive software. This trial will include both supervised and unsupervised phases. During the supervised phase, both groups will receive 40-minute sessions of supervised group training three times per week for a duration of 4 weeks. Participants will complete the first week of the intervention in groups of three at their local rehabilitation center with a trainer. The remaining 3 weeks will take place at participants’ homes using remote supervision by the trainer using Apple iPad technology. At the end of 4 weeks, the supervised period will end and the unsupervised period will begin. Participants will retain the Wii console and be encouraged to continue using the program for an additional 4 weeks’ duration. The primary outcome measure will be the “Two-Minute Walk Test” to measure walking capacity. Outcome measures will be evaluated for all participants at baseline, after the end of both the supervised and unsupervised phases, and after 1-year follow up.

Results: Study staff have been hired and trained at both sites and recruitment is currently underway. No participants have been enrolled yet.

Conclusions: Wii.n.Walk is a promising in-home telehealth intervention that may have useful applications for older adults with LLA who are discharged from rehabilitation or live in remote areas having limited or no access to existing rehabilitation programs.

Trial Registration: Clinicaltrial.gov NCT01942798; http://clinicaltrials.gov/ct2/show/NCT01942798 (Archived by WebCite at http://www.webcitation.org/6V0w8baKP).

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KEYWORDS

amputation; adult; aged; randomized controlled trial; telemedicine; walking

Introduction

In 2003, it was estimated that more than 2 million individuals were living with lower limb amputation (LLA) in North America with an annual incidence of 150,000 [1]. Over half of LLAs are transtibial (TT) and transfemoral (TF) amputations [2]. In Western countries, the incidence of LLA increases sharply after the age of 50 years as a result of secondary complications associated with illnesses such as diabetes and vascular disease [2].

Recovery following LLA is notably slow. A lengthy recovery process is especially common among older adults who often have multiple co-morbidities including peripheral vascular disease, peripheral neuropathy, hypertension, heart disease [3,4], and cognitive impairment [5]. LLA, compounded with these co-morbidities, influences walking and places these individuals at a high risk of falling and sustaining injury after a fall [6]. In fact, 52% of community-dwelling individuals with LLA report falling each year [7]. Similarly, 49% have a fear of falling and 65% report low balance confidence [7]. The consequences associated with these numbers may contribute to deterioration in balance [8], endurance, strength, and coordination [9] in older adults, and ultimately a decline in walking capacity. Walking capacity is a strong determinant of health-related quality of life (HRQOL) in individuals with LLA [10-13] as well as the best predictor of prosthetic walking in individuals with LLA [14]. The ability to walk longer distances allows the individual to move around his or her environment independently, which in turn impacts one's choice of activities and participation [15].

Following an LLA, individuals need to participate in prosthetic rehabilitation. Rehabilitation includes procurement of a prosthetic limb and ambulation training. The costs associated with post-amputation care and prosthetic rehabilitation are considerable. Post-LLA, projected lifetime health care costs total approximately US $509,000 [16]. Due to escalating costs, existing rehabilitation programs are experiencing difficulty providing sufficient levels of prosthetic therapy [1]. Furthermore, a trend toward outpatient rehabilitation, community inaccessibility, and transportation barriers imposes challenges for clients, particularly those in rural/remote areas, to attend face-to-face clinic appointments [17]. Therefore, accessible and innovative approaches are needed to improve outcomes for individuals with LLA and overcome these barriers to participation.

In-home telehealth is an innovative and emerging approach to provide rehabilitation through technologies and telecommunication [17-20]. Home treatments create accessible rehabilitation programs, promote continuity of care after discharge, and offset the time and expense of travel for clients to in-hospital rehabilitation programs [21,22]. Access to in-home rehabilitation is particularly important for those with limited access to facilities and transportation [23].

Nintendo Wii Fit is a commercial gaming technology that shows promise as an in-home rehabilitation tool. The benefits of using Wii Fit technology as a rehabilitation tool are demonstrated by the growing knowledge base on the use of gaming technology in older adult rehabilitation. In a study of older adults that used Wii Fit during in-patient rehabilitation, more than 80% expressed their desire to continue using Wii Fit at home [24]. Preliminary evidence suggests Wii Fit training is a feasible and safe method leading to improvements in balance [25,26], walking [25-27], and balance confidence in older adults [27]. Studies have reported improvements in walking and balance confidence in individuals with multiple sclerosis [28] and improved balance and decreased risk of falls in individuals with mild Alzheimer's [29]. Pilot testing has shown improvements in balance, balance confidence, and gait variables [25] in two older adults with LLA, which is consistent with findings from our own pilot work [30].

In a Single Subject Research Design (SSRD) study of six individuals with LLA, the feasibility of a Wii Fit oriented intervention consisting of structured daily training varying from 2 to 6 weeks was assessed. Results indicated a statistical improvement in walking capacity in five participants who had 3 or more weeks of intervention [30]. The aim of the present study is to extend findings on this topic and conduct a randomized controlled trial (RCT) to assess our in-home telehealth Wii Fit intervention protocol we call "Wii.n.Walk" (Clinicaltrial.gov NCT01942798).

The primary clinical hypothesis is that participants in the Wii.n.Walk intervention group will experience an improvement in walking capacity compared to the control group. The secondary clinical hypothesis is that participants in the Wii.n.Walk intervention group will experience an improvement in lower limb functioning (balance, gait speed, and strength), dynamic balance, physical activity, and balance confidence. The tertiary clinical hypothesis suggests that the Wii.n.Walk group will experience an improvement in life space mobility, prosthetic use, HRQOL, and will have a lower incidence of falls. The adherence hypothesis is that the Wii.n.Walk group will have ≥80% adherence.

Methods

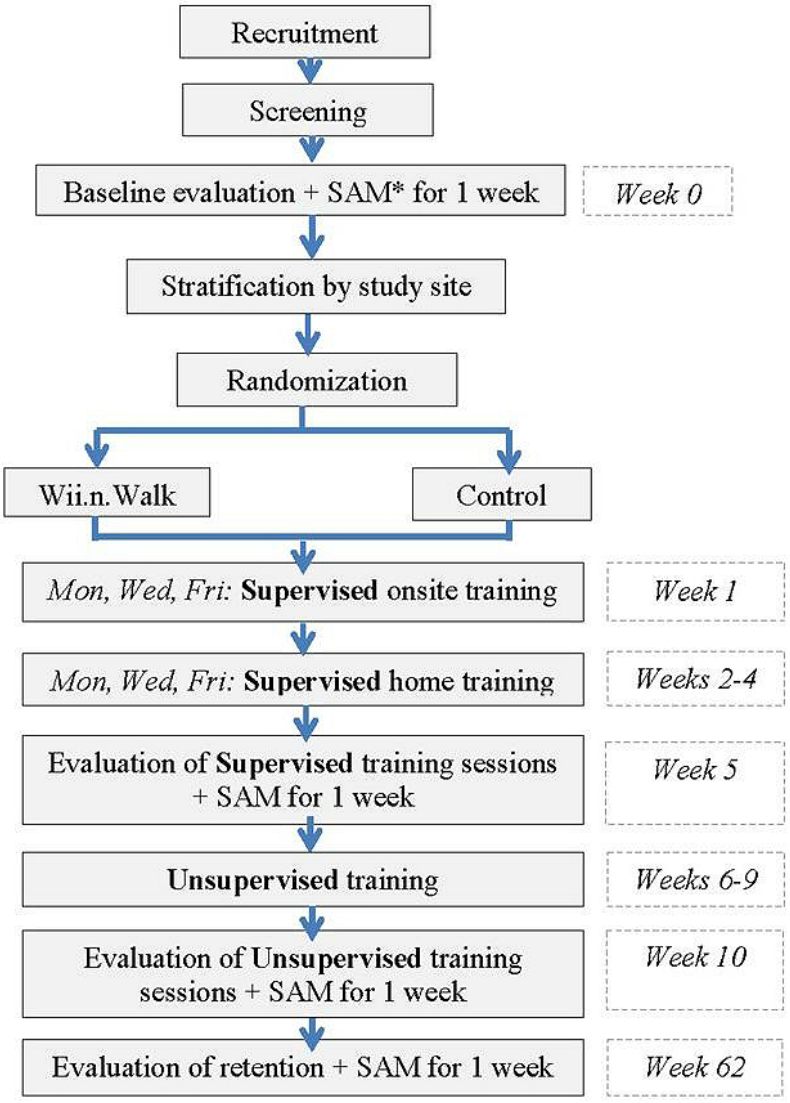
Trial Design

A parallel, evaluator-blind RCT conducted at two sites (Vancouver, British Columbia and London, Ontario) will be used. To minimize participants' bias associated with knowing which intervention is of interest to researchers, we will attempt to mask participants to the true study objectives (NCT01942798). This will be achieved thorough stating that "evidence suggests that having good cognition improves physical outcomes and we are trying to determine whether cognitive or activity training is better" both in the consent form and when addressing subjects' comments/questions.

Participants will be stratified by site and block randomized to the Wii.n.Walk intervention or control group in triplets using a 1:1 allocation ratio. To ensure balance between groups and masking of group assignment, a central computerized randomization process will be designed by the research team statistician, with undisclosed variable block sizes. Randomization will occur after the participant is screened and enrolled (Figure 1). The site coordinators will contact the statistician via telephone or email and obtain group assignment. The participant's contact information will be forwarded to the appropriate group trainer to arrange for an initial training session.

This trial includes a supervised and an unsupervised phase for both the Wii.n.Walk intervention and control group. Once enrollment has been completed, participants will be evaluated on a number of clinical measures at baseline, after the supervised phase is completed, after the unsupervised phase is completed, and at a 1-year follow-up time point.

Figure 1. Study Flowchart.



Participants

A total of 72 community-dwelling prosthetic ambulators in London and Vancouver will be recruited through clinicians and prosthetists. A letter of information will be distributed to all individuals in the amputee program databases who meet the study inclusion criteria. Participants need to be ≥50 years of age, have a unilateral TT or TF amputation, use their prosthesis for at least 2 hours per day for the past 6 months to minimize the influence of residual limb/prosthetic fit problems, be

cognitively able to engage in the program (receive a score on the Modified Mini-Mental Status Exam score of >23) [31], and have a television that will enable connection to the Nintendo hardware.

Individuals will be excluded if they cannot communicate in English, cannot provide informed consent, have medical conditions (eg, congestive heart failure) that limit exercise participation as determined using the American College of Sports Medicine exercise guidelines for older adults [32], have prosthetic fit issues (eg, pain and discomfort) as indicated by scores <6 on the Prosthetic Socket Fit Comfort Scale [33], or are currently participating in another supervised exercise or training program (eg, balance training).

Participants between 50-69 years old will be medically screened by the site coordinators using the Physical Activity Readiness Questionnaire (PAR-Q) [34]. Participants who are 70 or more years old or who answer "yes" to any of the PAR-Q questions will be medically screened by a physician using the Physical Activity Readiness Medical Examination (PARmed-X) to obtain clearance for physical activity participation [35].

Wii.n.Walk Intervention

Participants in the experimental group will receive the Wii.n.Walk intervention. The Wii.n.Walk intervention was developed by core members of the research team and refined based on observations and the feedback received from the participants in the pilot studies [30]. Modifications for trainer instructions were made to Wii Fit postures and activities to prevent incorrect postures/techniques and to promote function and safety. Preliminary work also informed the dosage/frequency and duration of the intervention.

Social Cognitive Theory (SCT) [36] is the theoretical foundation for the Wii.n.Walk intervention. This theory was developed to enhance all four sources of self-efficacy: performance mastery, vicarious learning, verbal persuasion, and reinterpretation of physiological responses. Performance mastery, or learning to perform a specific skill, is the most robust source of self-efficacy. Successful performance of the Wii.n.Walk activities may provide a sense of accomplishment and thereby improve self-efficacy. Vicarious learning, or learning by watching others successfully accomplish activities, provides the observer a sense that they, too, have the ability to accomplish the task. This will be established by performing the Wii.n.Walk activities in groups initially and having participants watch the other group members perform the activities. Verbal persuasion will arise from credible feedback, guiding the learner through the task, and motivating his or her best effort. The trainer will provide this feedback when appropriate—at least once each session for each group member. The Nintendo device also automatically provides auditory and visual feedback based on the participant's performance. Finally, participants will be taught to reinterpret physiological responses (eg, stress and anxiety) that may be associated with challenging Wii.n.Walk activities. According to a systematic review, physical activity programs that incorporate SCT are more effective in enhancing adherence [37]. More specifically, social support, peer modeling, and group training have been identified as important factors for increasing adherence in older adults [38].

The Wii.n.Walk protocol consists of Wii Fit activities. Participants stand on the Wii Fit balance board and play the games through weight shifting or by using the Wii remote control. The intervention protocol includes selected activities and exercises including yoga (static and dynamic single and double leg poses), balance tasks (lateral, posterior, and anterior weight shifting exercises), strength training (dynamic single and double leg exercises), and aerobics (running on the spot and step class).

Participants will complete two training phases: a 4-week supervised phase (3 times/week, 40 minutes training /session) followed by a 4-week unsupervised phase. Based on our pilot studies, completion of 40-minute Wii Fit exercises in each session takes approximately 60-90 minutes for the participants, including the instructions given by the Nintendo software as well as the rests that may be needed between the exercises. Participants will meet in groups of three at their local rehabilitation center with an experienced trainer during the first week to learn the program. They will then complete the final 3 weeks of the supervised program at home while being remotely supervised by the trainer and remotely interacting with the other two group participants. During the supervised phase, the trainer will provide individualized intervention and advance the training as the participant improves. At the end of the 4-week supervised phase, participants will retain the Wii units and be encouraged to use the program on their own for an additional 4 weeks (unsupervised phase).

The supervised phase purposely begins with in-person visits to introduce participants to the program, initiate group dynamics (eg, peer support), and create familiarity with the activities in a monitored safe environment. Graduating to home sessions overcomes barriers, cost, and inconveniences associated with travelling to a rehabilitation clinic and is intended to reinforce continued participation [22]. Supervised phase home sessions will be monitored by a trainer remotely using iPads with Wi-Fi plus cellular (Apple Inc, Cupertino, California, USA), preloaded with the VidyoMobile videoconferencing application (Vidyo Inc, Hackensack, New Jersey, USA). VidyoMobile enables the participant to meet at home with the trainer and the other two participants in the group. For better sound quality, participants will be asked to wear wireless headphones (Kinivo, Bellevue, Washington USA) with noise cancellation features. The iPad interface has been simplified as much as possible. Only the VidyoMobile app is available to the participants (access to all other apps is disabled through the iPad's parental control feature), and they can connect to the trainer by entering only their names and a simple PIN code. iPads will be securely mounted on a sturdy tablet tripod and will be placed a few meters away from the participant's TV and behind the participant, so that the trainer can see both the participant's screen as well as his or her posture. The ideal location will be established by the trainer during the home setup of the Wii.n.Walk equipment. At the beginning of each session, the trainer will hold a brief discussion session with all participants, through the videoconferencing software, to review the plan for the session and address any questions. Once the session begins, the trainer is able to watch and supervise all three participants from his or her desktop/laptop at the clinic. The participant's

iPad can display the trainer and the other two participants. The trainer can remotely deactivate each iPad's camera to reduce distractions while exercising or to reduce video streaming costs. The trainer will activate the iPad's camera on at least two occasions during each session to enable opportunities for vicarious learning and participant-to-participant verbal persuasion. As an example, the trainer will ask two participants to watch the third perform an exercise. Verbal persuasion will be provided by the trainer to the participant being watched.

The exercises/games and their difficulty levels will be chosen by the trainer. The three participants in the same group will perform similar sets of exercises/games; however, the difficulty level of the exercises/games will vary depending on each participant's abilities. By default, the more challenging levels of the games are initially locked and can only be unlocked if the participant successfully completes easier, prerequisite levels. In addition, progression to more difficult and longer activities is guided by instructions in the Wii.n.Walk manual. The manual also provides modified activity positions such as adding unilateral or bilateral external hand support if required by the participant. Modifications can be made if the participant has difficulty or is unable to do the activity. As an example, activities may be modified for an individual with a TF amputation if the prosthesis is not structurally capable of assuming the exercise position (eg, some of the exercises require stance phase prosthetic knee flexion). Common postural mistakes are included in the manual to guide the trainer in correcting positioning.

At the end of the supervised phase, the iPads, stands, and headphones will be collected by the trainer, while participants will retain the Wii console and balance board for the unsupervised phase. Participants will be encouraged to use the Wii.n.Walk program as much as they like during the unsupervised phase by continuing to do the same exercises/games they did during the supervised phase and progressing to the challenging levels if they unlocked those levels. To avoid confounding information, other people living with the participant will be asked not to use it. The trainer will telephone the participant once a week to monitor for safety (eg, falls) and equipment function.

For both supervised and unsupervised phases, depending on the level of ability and potential for safety issues, participants will be asked to have two high-back chairs placed on either side of them to minimize the risk of falling while using the Wii.n.Walk program. For participants who require additional assistance, if available, a family member, friend, or caregiver will be encouraged to be present during the training sessions. As the participant's abilities improve, they will progress from the use of such assistance.

Control Intervention

The control group will follow the same protocol but will be trained to use the Wii Big Brain Academy Degree program (Nintendo, Kyoto, Japan). Big Brain is a low-cost, commercially available software consisting of video games to improve cognitive function. Participants will use the Wii remote to participate in the games by pointing and clicking to select answers in response to on-screen questions. Big Brain games require participants to identify, memorize, analyze, compute, and visualize. The games have easy, medium, and difficult levels. Participants initially start with easy games and progress to more challenging levels based on their performance. The trainer will design the supervised sessions, provide instruction/feedback, and facilitate group discussions. Results from the feasibility study indicate that participants enjoyed discussion about topics including which games are harder, strategies for doing better at different games, and comparing scores.

We chose cognitive video gaming for the control intervention because (1) it enables non-specific attention control, (2) there is minimal concern that it will impact the primary outcome because of its non-physical nature, (3) it uses similar technology as Wii.n.Walk, (4) our feasibility data suggest that it maintains motivation and therefore decreases attrition, and (5) it is potentially beneficial and ethically acceptable [39]. Two separate trainers will administer the Wii.n.Walk and control interventions to minimize treatment bias.

Outcome Evaluation

Overview

Outcomes will be evaluated (Figure 1) by blinded evaluators at baseline, end of the supervised phase (week 5), end of the unsupervised phase (week 10), and end of retention period (week 62). The evaluators will be senior university students in health sciences and will have at least 1 year of experience working with research participants. They will be trained by co-investigator, BI, who has more than 5 years of experience working with amputees and administering the outcome measures used in this study.

Primary Outcome Measure

The Two-Minute Walk Test (2MWT) will be used to measure walking capacity as the primary outcome measure. Starting from a standing position, participants will be asked to walk as far as they can in a safe manner for 2 minutes over an indoor, flat, out-and-back course. The distance travelled to the nearest meter is recorded. The Canadian Physical Medicine and Rehabilitation Association's Amputee Special Interest Group [40], and others [41,42], have recommended the 2MWT as the preferred measure of walking capacity. It is used in more trials of individuals with LLA [43-53] than any other measure, enabling us to compare our results with previous studies. The 2MWT has been validated with a number of LLA samples [41,42,51-53]. The 2MWT has demonstrated intra-rater reliability (intraclass correlation coefficient/ICC=.96), inter-rater reliability (ICC=.98) [51], and validity and responsiveness to change (mean 13.6, SD 19.9 meters) in individuals with LLA [52].

Secondary Outcome Measures

The Short Physical Performance Battery (SPPB) will capture timed standing balance (parallel foot stance, semi-tandem, or tandem: 10 seconds each), lower limb strength captured using time (to the nearest second) taken to complete five sit-to-stand chair transfers (no hand support), and gait speed (to the nearest second) over 4 meters using a standing start [54]. There is

support for test-retest reliability (ICC=.92) and validity in older adults with disability [55,56]. Due to observing a ceiling effect for this measure in our earlier pilot work, we modified the scoring of the scale by timing each of the standing balance tasks for up to 30 seconds. An additional item, timed single leg stance (up to 30 seconds for each leg), was added to evaluate single leg stance balance.

The Four Square Step Test (FSST) will be used to measure dynamic standing balance. Electric tape is used to create four squares on the floor [57]. The participant is asked to step in each square, first clockwise and then counter-clockwise, without touching the tape, as fast as possible, and with use of his or her walking aid if needed. This test is timed and faster times indicate better dynamic standing balance. Scores ≥24 seconds indicate the individual is at risk for falls [58]. FSST has shown to be reliable (ICC=.98) and valid in older adults [57].

The Physical Activity Scale for the Elderly (PASE) is a self-report measure that captures information on the frequency, duration, and intensity of various physical activities [59]. The 10-item PASE has two parts: Part 1, Leisure Time Activity, has six items about involvement in daily activities such as participating in light exercise during the past 7 days. The response options are “never”, “seldom”, “sometimes”, or “often”. Information on the type and the mean time spent engaging in the activity per day is also captured. Part 2, Household Activity, has three “yes/no” items about participation in daily activities. The last question asks about number of hours per week, as well as the amount of physical activity involved, in paid or volunteer work. The amount of time spent and participation (yes/no) are multiplied by a weighted value. The total PASE score is derived by summing each contribution and varies from 0 to 500, with higher scores representing higher physical activity levels. Test-retest reliability (ICC=.84) and validity have been reported for older adults [59].

The Activities-specific Balance Confidence (ABC) is a 16-item self-report scale to assess perceived balance confidence [60]. The item scores are summed and divided by 16 to derive a mean overall score varying from 0 to 100, with higher scores indicating more confidence. Validity and test-retest reliability (ICC=.91) have been shown in individuals with LLA [61].

Tertiary Outcome Measures

Life Space Assessment (LSA) is a 5-item scale that will be used to measure the size of the spatial area that an individual moves through in his or her daily life, as well as the frequency of his or her mobility within a certain timeframe [62]. Life space level (where participants travel) is measured dichotomously (yes/no), frequency is measured on a Likert Scale (1 to 4) from less than once/week to daily, and independence is measured in terms of the need for aids or equipment or assistance from another person. The total score for each item is the product of the life space level, frequency, and independence. All items are summed for a final score. Evidence for validity and test-retest reliability (r=.86) has been reported for older adults [63].

Modus Health Stepwatch Activity Monitor (SAM) will be mounted on the prosthetic ankle to record number of steps taken per time interval to indicate the amount of prosthetic use (Modus Health, Washington, DC). The SAM cannot be adjusted by the participant and needs to be connected to a computer with special software for programming and data downloading. It has a 99.4% accuracy in individuals with LLA for a wide range of gait styles, from slow shuffle to a fast run [64,65]. The SAM will be used to collect data in 1-week intervals at all evaluation times.

Health Utility Index Mark 3 (HUI3) is useful in performing cost-utility and cost-effectiveness analyses of new rehabilitation interventions. The HUI3 is a brief questionnaire about health status reflected in a measure of HRQOL [66]. Each single-attribute utility is scored between 0.00 and 1.00 and the multiple-attribute utility scale is scored from −0.36 to 1.00, with higher scores reflecting better health and quality of life. Test-retest reliability (ICC=.72) has been shown in patients recovering from hip fracture [67]. Differences of 0.03 have been found to represent meaningful change [68]. This study is not sufficiently powered to undertake a cost-utility analysis, but it will provide useful utility data to estimate what changes in HRQOL might be anticipated [68].

The Walking While Talking Test (WWT) is a test of divided attention to examine cognitive-motor interactions [69-72]. The WWT requires the ability to divide and switch attention between two tasks, and it has been reported that older adults show an innate preference for preserving gait over talking [73,74]. Participants walk 6 meters on a flat course, turn around, and walk 6 meters back to the start while reciting the letters of the alphabet (a, b, c, ...) aloud (WWT-simple). They repeat this routine while reciting alternate letters of the alphabet (a, c, e, …) aloud (WWT-complex). The difference in time (to the nearest second) to complete the simple and complex walks will be calculated with higher differences suggesting poorer ability to cope with dual tasks (eg, greater need to focus on walking). Inter-rater reliability (r=.602) and validity have been reported in older individuals [75]. The WWT will be collected with the goal of “misdirecting” participants and masking the study objectives.

The Fall Calendar will be used to document the number of falls, circumstances (eg, cause, location, assistive device used or not), and consequences participant have had (eg, medical visit, injury) over the course of study.

Adherence will be measured by total amount of the program use (minutes, frequency, and duration), which will be collected from the Wii console at the time of equipment pick-up.

Primary Analysis

To account for any within-cluster correlation that may occur as a result of delivering the intervention in groups, ICC will be calculated among the outcomes of participants within the same groups (clusters). Variation inflation factor (VIF) will be calculated using the formula: VIF=1+ICC(M−1) [76-78]. The M variable refers to the cluster size, which equals 3 in this study. Post-treatment walking capacity scores will be compared in the Wii.n.Walk and control groups using analysis of covariance (ANCOVA) for the end of supervised phase, end of unsupervised phase, and end of retention period, controlling for site, baseline score, and possibly amputation level and age [79]. To adjust for clustering effect, the ANCOVA’s F statistics will

be divided by the VIF [76,77]. Missing data will be handled using Multiple Imputation [80]. A sensitivity analysis will also be conducted to evaluate the impact of missing data [81]. Significance testing (*P*) and marginal means with 95% confidence intervals will be estimated. Effect size (partial eta squared) will be calculated as a ratio of the effect and total sums of squares, with a 95% confidence interval. Primary analysis will be based on intention-to-treat to include all randomized participants. However, secondary analysis on a per-protocol basis (participants who adhere to treatment) will also be conducted for comparison [82].

Secondary/ Tertiary Analyses

ANCOVA with an adjusted *F* statistic (as explained above) will be used to compare post-treatment scores between groups for secondary and tertiary outcomes. Confidence intervals (95%) will be derived. Mean percent adherence will be calculated.

Sample Size Calculation

The primary outcome (2MWT) was used to calculate the sample size. The responsiveness of the 2MWT in a single study that included older adults with LLA being discharged from rehabilitation had a mean of 13.6 (SD 19.9) meters [52]. In an RCT of younger (mean age 36 years) community-dwelling individuals with LLA, the mean difference between treatment and control groups was 11.2 (SD 18.4) meters [43]. Walking distance gains from our own pilot work (mean 25, SD 18.1 meters) on younger adults in rehabilitation [30], and early results from our feasibility study (mean 11.9, SD 9.3 meters) using a similar sample as the proposed study, suggests distances of up to 14 meter gains may be possible. Taking into account these data and based on our study team's considerable clinical expertise, we decided that the minimal clinically important difference of 14 meters would be reasonable. Using the sample size calculation formula for ANCOVA in RCTs (α=.05; β=.1; rho=.72) [82], each group would require 21 participants. Borm et al [83] demonstrate that when rho lies between .2 and .8, ANCOVA further reduces the required sample size by 10-40% over change score. Accounting for an additional 4 degrees of freedom (one stratification factor for site at randomization and possibly three at analysis), an extra four participants per group (n=25) are required. Accounting for a conservative total group dropout rate of 25% and supported by physical activity clinical trials (8-24%) [84,85], 64 participants are required. Because the supervised sessions are conducted in groups of three, the sample size needs to be divisible by three. Therefore, a final n=72 will allow us to enroll a balanced n of 18 Wii.n.Walk and 18 controls at each site.

Safety

The Wii.n.Walk manual incorporates extensive safety-related material, including teaching correct postures and avoiding unsafe situations. Any unsafe performance observed during training will be addressed immediately with corrective feedback. Participants will be instructed to regularly report their Perceived Rate of Exertion (PRE) rating from 6 to 20 (6=no exertion at all to 20=maximal exertion) to exercise in a safe zone [86]. They will also be asked to report on their pain and fatigue on a scale of 0 to 10 (0=no pain/fatigue to 10=worst possible pain/fatigue) during the session. Participants will be asked to stop the session if their PRE level is ≥14 (ie, hard or heavy PRE) or fatigue and pain levels are ≥7. For the unsupervised sessions, participants will be advised to self-monitor their own safety using the PRE and pain/fatigue scales. Participants will be encouraged to contact the site coordinator immediately if they experience any unusual discomfort, pain, or physical symptoms.

There will also be a foot/leg assessment that will be completed with the participants. At all evaluation points and at enrollment, the co-investigator, BI, or the research coordinator, will ask participants if, since their last assessment (or if at enrollment session, in the last 2 weeks), they have experienced any of the following new problems in their non-amputated and amputated limbs: skin irritation/open sores/wounds, pain, swelling, or other medical problems that have prevented/stopped them from using their prostheses as they normally would. If the participants say that they have noticed issues, BI, or in London, the research coordinator, will check the participants' foot and stump. If BI or the research coordinator notices any issues, he or she will refer the participants for medical assessment by their medical doctor or the study doctor.

A Data and Safety Monitoring Board (DSMB) will review accumulating outcome data and advise the investigators regarding safety issues, evidence of benefit, and need for modification to the study design. The DSMB will include three members external to the research team: a statistician, a prosthetist, and an older adult with LLA. Adverse events (eg, injury, falls) will be documented by the trainer and reported to the DSMB and the applicable Ethics Review Board.

Ethics and Funding

This study has been approved by the Ethics Boards at the University of British Columbia (Approval #: H13-01858) and Western University (Approval#: 104688), as well as the Research Review Committee for the regional health authority at each site. This study is funded by the Canadian Institutes of Health Research (CIHR) [MOP-130336], a grant from the Amputee Coalition of Canada, and the University of Alberta-Franklin Fund. BI is a Vanier Canada Graduate Scholar.

Results

Study staff have been hired and trained at both sites and recruitment is currently underway. No participants have been enrolled yet.

Discussion

Overview

The lack of continuity in rehabilitation treatment after discharge leads to decreased mobility, walking capacity, functional independence, and HRQOL. Wii.n.Walk presents a promising in-home telehealth intervention that may have useful applications for older adults with LLA who are discharged from rehabilitation, live in remote areas, or have limited or no access to existing rehabilitation programs. The application of a home treatment intervention reduces the client's burden for travel and is particularly advantageous for clients with greater disability.

The benefits extend beyond improved walking capacity and include increases in physical activity tolerance and better health and participation in important life activities that may ultimately ameliorate social and financial costs.

Limitations

This study has a number of limitations. RCTs in rehabilitation are subject to numerous threats to study validity [87]. First, blinding is a difficult limitation to overcome. Evaluators will be blinded and will request the participants not reveal their group status. We will also endeavor to mask participants to the true study objectives. It still may be evident which intervention is of primary interest based on the outcomes used. Therefore, we attempt misdirection by including the WWT test and by stating that "We are trying to determine whether cognitive or activity training is better for improving function" both in the consent form and when addressing participants' comments/questions. Second, contamination and co-intervention will be difficult to control because the Wii Fit and the Big Brain are commercially available. While masking study objectives may reduce these risks, we will also conduct the in-clinic sessions at different times during the day so that participants will not have contact with the other group. Third, although the trainers will ask the participants not to use the treatment software outside of the treatment schedule, they may ignore these requests. Date/time stamped Wii use data downloaded at the end of intervention from the consoles will be assessed to determine adherence. This will also enable us to explore if individuals other than the participant used the Wii. Fourth, not everyone likes video games; however, the pilot work and the Wii Fit literature [72] suggest that the majority of older participants enjoy the games. Fifth, there is a possibility for a technology burden, particularly for older participants. We endeavored to simplify the technology used in this study to minimize the burden. We will use color-coded dots to highlight important buttons on the iPads, headphones, and the Wii consoles, so it will be easier for participants to locate them. Participants will be trained on how to use each piece of the technology during their in-clinic sessions as well as during the equipment set up at their homes. They will also be provided with a take-home manual that clearly explains step-by-step guidelines for using the program. In case participants have difficulty connecting with the trainer during the supervised phase, the trainer will troubleshoot remotely by telephoning participants. Sixth, our sample will represent older adult volunteers. The results will not be generalizable to younger amputees. We do not view this as a limitation given that >80% of individuals with LLA in Western countries are older adults. The results will be limited to differences related to older volunteers. Finally, loss to follow-up is a threat, particularly when participants are based remotely. To minimize loss to follow-up, the site coordinators will maintain contact with participants once a month upon termination of the unsupervised phase under the premise of collecting information on falls.

Acknowledgments

WCM was responsible for overall administration of the grant, design, and oversight of the study, and site-lead for Vancouver. JJE provided guidance with the research design and implementation. IMM was instrumental in the development of the technology adaptations for the Wii.n.WALK and provided ongoing technology design and support. CHG provided expertise in the RCT design and analysis as well as cost-utility and cost-effectiveness evaluation. HF provided expertise in terms of the design of the Wii.n.Walk and the choice of outcome measures. MWCP is the site-lead for London and contributed to the conception and development of the study. TJ contributed to the design of the Wii.n.Walk related to gaming applications. BI was primarily responsible for development of the Wii.n.Walk, pilot testing, and writing the first draft of the manuscript. All authors contributed to and reviewed the final version.

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Conflicts of Interest

None declared.

Multimedia Appendix 1

CONSORT-EHEALTH checklist V1.6.2 [88].

[PDF File (Adobe PDF File), 1MB - resprot_v3i4e80_app1.pdf]

Multimedia Appendix 2

WiiNWALK 2013 CIHR peer review report.

[PDF File (Adobe PDF File), 126KB - resprot_v3i4e80_app2.pdf]

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Abbreviations

2MWT: 2 Minute Walk Test
ABC: Activities-specific Balance Confidence
ANCOVA: analysis of covariance
CIHR: Canadian Institutes of Health Research
DSMB: Data and Safety Monitoring Board
FSST: Four Step Square Test
HRQOL: health- related quality of life
HUI3: Health Utility Index Mark 3
ICC: intraclass correlation coefficient
LLA: lower limb amputation
LSA: Life Space Assessment
PARmedX: Physical Activity Readiness Medical Examination
PAR-Q: Physical Activity Readiness Questionnaire
PASE: Physical Activity Scale for the Elderly
PRE: perceived rate of exertion
RCT: randomized controlled trial
SAM: Stepwatch Activity Monitor
SCT: Social Cognitive Theory
SPPB: Short Physical Performance Battery
SSRD: single participant research design
TF: transfemoral
TT: transtibial
VIF: variance inflation factor
WWT: Walking While Talking Test

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Protocol

Improving Outcomes in Cancer Patients on Oral Anti-Cancer Medications Using a Novel Mobile Phone-Based Intervention: Study Design of a Randomized Controlled Trial

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Abstract

Background: The widespread and increasing use of oral anti-cancer medications has been ushered in by a rapidly increasing understanding of cancer pathophysiology. Furthermore, their popular ease of administration and potential cost savings has highlighted their central position in the health care system as a whole. These facts have heightened appreciation of the unique challenges associated with the use of oral anti-cancer medications; especially in the long-term use of these medications and the associated side effects that may impede optimal adherence to their use. Therefore, we developed ChemOtheRapy Assistant, CORA, a personalized mobile phone–based self-management application to help cancer patients on oral anti-cancer medications.

Objective: Our objective is to evaluate the effect of CORA on adherence to oral anti-cancer medications and other clinically relevant outcomes in the management of patients with renal and prostate cancer.

Methods: The study will be implemented as a 2-parallel group randomized controlled trial in 104 patients with renal or prostate cancer on oral anti-cancer medications over a 3-month study period. The intervention group will use CORA in addition to usual care for self-management while the control group will continue care as usual. Medication adherence will be measured objectively by a Medication Event Monitoring System device and is defined as the percentage of prescribed doses taken. We will also assess the effect of the intervention on cancer-related symptoms measured by the MD Anderson Symptom Inventory and unplanned hospital utilizations. Other outcomes that will be measured at study start, midpoint, and endpoint are health-related quality of life, cancer-related fatigue, and anxiety. Group differences in medication adherence will be examined by t tests or by non-parametric Mann-Whitney tests if the data are not normally distributed. Logistic regression will be used to identify potential predictors of adherence.

Results: We expect to have results for this study before the end of 2016.

Conclusions: This novel mobile phone–enabled, multimodal self-management and educational intervention could lead to improvements in clinical outcomes and serve as a foundation for future mHealth research in improving outcomes for patients on oral anti-cancer medications.

(JMIR Res Protoc 2014;3(4):e79) doi:10.2196/resprot.4041

KEYWORDS

cancer; oral anti-cancer medication; mobile application; randomized controlled trial; self-care; mHealth; medication adherence

Introduction

Oral anti-cancer medications (OAMs) are increasingly being used as an alternative to traditional intravenous chemotherapy in cancer management [1]. Factors promoting this new trend in cancer management include increased survival times requiring long-term therapy, favorable acceptability of these newer medications among patients, convenience and ease of administration, and potential cost savings due to less time spent in the hospital [2,3]. However, this shift significantly increases the burden of self-care for patients similar to that in many chronic diseases where compliance, defined as "implementation by the patient of the therapeutic plan that has been established", is the direct responsibility of the patients or caregivers [3]. Medication adherence in cancer therapy has significant impact on treatment efficacy and development of toxicities [4]. Studies have reported variable medication adherence rates in cancer patients on OAM regimens, with adherence rates ranging from less than 20% to 100% [5,6]. Suboptimal adherence not only leads to loss of treatment efficacy and increased toxicity, but it also results in increased hospital utilization, longer hospital stays, and increased health care costs [6,7].

An important component of self-managing these medications is managing the adverse effects, which are not altogether different from their intravenous counterparts, resulting from taking these powerful medications. These adverse effects threaten their continued use, and most commonly result in non-adherence to the recommended treatment plan [8]. In addition to the drug-related adverse effects, other barriers to optimal adherence include inadequate treatment supervision, poor communication with providers, and other patient-related barriers like poor knowledge, self-efficacy, and motivation [9]. Multidisciplinary approaches to home-based management of chemotherapy by oncology nurses, physicians, and health educators as well as some pharmacy-based care management programs have shown some success in addressing this problem [10-12]. However, these approaches are not scalable unless they are made more cost-effective. We hypothesize that creatively leveraging mobile technology could potentially mimic the effect of such intensive and coordinated interventions to improve outcomes.

Therefore, we developed ChemOtheRapy Assistant, CORA, a personalized mobile-based, multimodal self-management intervention designed for extensive patient education and symptom management, based on evidence from clinical guidelines and direct patient/caregiver stakeholder feedback, to improve medication adherence in cancer patients on OAMs. We expect that this intervention, delivered through a mobile device, compatible with both iOS and Android devices, will forge engagement to improve outcomes similar to those seen in chronic disease management programs [13-15]. This novel project is being implemented in a collaborative effort by a multidisciplinary team of oncology physicians, nurses, pharmacists, and psychologists.

For this study, we will focus on patients with renal cell cancer (RCC) and prostate cancers. These specific cancers accounts for significant morbidity and mortality due to cancer in the United States [16]. OAMs are either first-line therapies (eg, for RCC) or very commonly used at some point in management of these cancers; and with increasing survival data, there is evidence for their use on an extended basis. We will be including patients with RCC or prostate cancer on any of the following four oral VEGF inhibitors—sorafenib, sunitinib, pazopanib, axitinib—or everolimus, an mTOR inhibitor. These OAMs are commonly used in patients with metastatic renal cell carcinoma with common side effects including hypertension, diarrhea, nausea, and hand-foot syndrome [17]. We hypothesize that patients with RCC and prostate cancers on OAMs who use CORA will develop increased competency for self-care to manage side effects associated with their medications, which will translate to higher medication adherence and improved clinical outcomes compared to cancer patients on OAMs who do not use the mobile-based intervention.

Our primary aim is to assess the effect of CORA on adherence to OAMs. We define medication adherence as the percentage of prescribed doses taken.

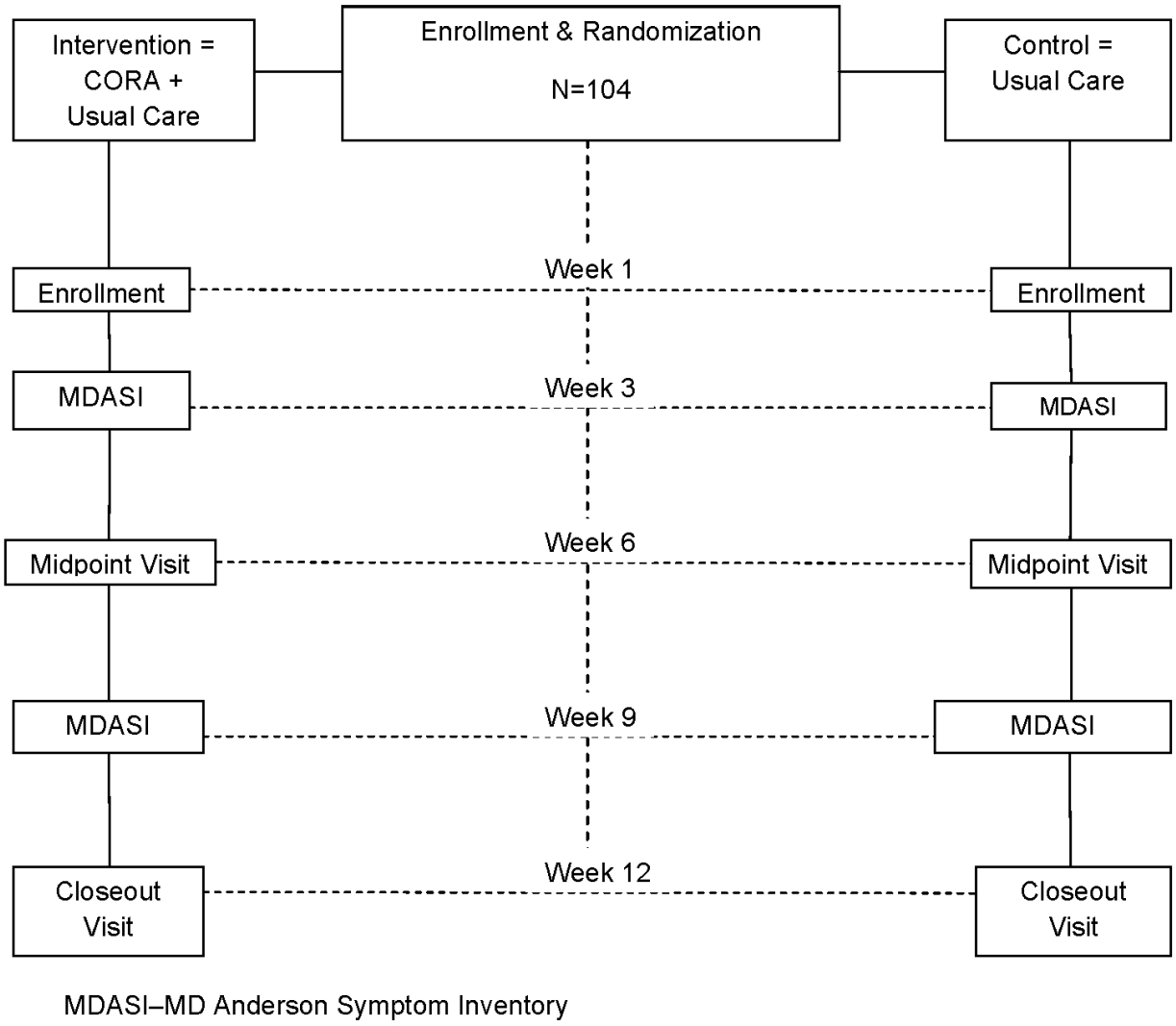
For our secondary aims, we will be assessing the effect of CORA on the severity of symptoms, unplanned hospital utilizations, health-related quality of life, cancer-related fatigue, and anxiety in patients on OAMs. Additionally, we will also assess usability and participant satisfaction with the app.

Methods

Trial Design

This study will be implemented as a 2-parallel group randomized controlled trial, intervention versus usual care, with multiple assessments over a 3-month follow-up period. Figure 1 shows the research design.

Figure 1. Schematic summary of the trial design.



Participant Inclusion/Exclusion Criteria

Participants for this study will be recruited from the Dana Faber Cancer Institute (DFCI), Boston, Massachusetts. To be considered eligible to participate in the study, patients must fulfill all eligibility requirements: patients must be ambulatory, age 18 years or older, able to consent for self, have a diagnosis of renal or prostate cancer, commencing a new cycle of OAMs, and able to read and speak English. In addition, since the intervention will be deployed on a mobile phone, they must have an Apple or Android mobile phone and be willing to download the app on their mobile phone to use the intervention.

Exclusion criteria include life expectancy less than 3 months as determined by the managing oncologist, current participation in a similar study geared at improving medication adherence or in investigational drug trials where adverse effects have not been fully elucidated, and presence of significant psychiatric comorbidities and memory or cognitive impairments. A significant psychiatric condition includes any condition that creates major distress for a patient or markedly impairs the patient's daily functioning. This includes, but is not limited to, acute psychoses, major depressive disorder, dementia, etc.

Recruitment Procedure

Participants for this trial will be drawn from cancer patients receiving care at the DFCI on OAMs. On a weekly basis, the research team will identify patients being started on a new cycle of OAM from chart reviews. After identifying potentially eligible patients, we will ask the managing oncologists to give approval to contact the patient about participating in the study. Thereafter, recruitment letters will be sent to all approved patients. One week after recruitment letters have been mailed, research assistants will attempt to contact subjects by phone to provide further detail regarding the study. If subjects are interested in participating, study staff will schedule an enrollment visit with subjects at the DFCI at the subject's convenience.

All enrollment procedures, including the informed consent process, completing enrollment surveys, and randomization procedures will be performed by trained research assistants. At the enrollment visit, all participants will be instructed to continue to receive usual medical care from their physicians as usual. They will also receive a Medication Event Monitoring System (MEMS) device, a valid measure of adherence used in many chronic diseases, to monitor medication adherence in this study. The bottle cap has a microprocessor that records all instances

and times that the bottle is opened. They will be instructed to open their pill bottles only when they want to take their medications. Only subjects randomized to the intervention arm will download and be able to use the CORA mobile app.

Intervention

Framework for Intervention

This mobile-based multimodal self-management program is based on integrated evidence from the American Society of Clinical Oncology and the Oncology Nursing Society standards for safe chemotherapy administration clinical guidelines, the medical literature, and experiences from clinical practice. Our goal is to increase adherence to OAMs by empowering patients to better manage their medications and associated side effects. The focus will be on helping patients develop competency in preventing, or the early identification of, adverse effects that may impact adherence and hence negatively impact clinical outcomes. Our approach is grounded in extensive education and symptom management. Our strategies are as described below.

Coaching to Improve Self-Efficacy for Self-Care

Prior to the onset of oral chemotherapy, intensive education (60-90 minutes in duration) is usually done in the hospital at a time when patients are anxious and distracted by the high volume of information being directed at them. Additional educational sessions are also done on a monthly basis or prior to the commencement of a new treatment cycle. Instead of this intensive episodic education, a more frequent, bite-size coaching strategy will be adopted. Participants will receive daily push notifications from the educational library of CORA. This strategy is focused on empowering patients to be able to self-manage the recommended home care activities. The content of the messages include:

- general education to improve patients' understanding about the disease process
- education for patients about their medications and associated side-effects
- information about the benefits of optimal adherence
- strategies to help prevent or delay side-effects
- education about coping and self-care skills
- psychosocial support
- information about safe handling, storage, and best practices in taking medications
- information about safety measures in disposing of bodily waste products and addressing other safety issues pertaining to the use of chemotherapeutic medications at home

Symptom Reporting and Management

Since patients have to take medications in the comfort of their homes without their oncology nurses or physicians to monitor or manage side effects, they need to be able to identify side effects early in order to institute management in a timely fashion. Symptom management is based on three premises that patients can:

1. Recognize symptoms at an early stage. Patients will receive educational messages, about common symptoms that accompany their OAMs such that they are able to identify these symptoms early and manage or report them in a timely fashion.
2. Assess severity of their symptoms. After patients have been able to identify their symptoms, they will be able to use the mobile app to assess the severity of their symptoms, after which they will be guided on appropriate management strategies.
3. Generate appropriate response to the symptom. Response to symptoms can be negative or positive. A common negative reaction is to ignore symptoms that may become self-perpetuating and eventually result in decreased medication adherence or increased toxicity. The positive response we aim to help patients develop is increasing competency at performing basic self-care activities so that in time, patients become experienced managers of these common symptoms during a repeat episode.

Existing symptom management protocols at the DFCI will serve as the framework of the symptom management algorithm. After symptoms have been identified and adequately quantified, the app will guide patients through self-care strategies to help them adequately manage the symptoms.

Symptoms are categorized into red-flag symptoms (Figure 2) that notify the patients' care providers and other symptoms that can be managed through the app. Participants will receive weekly prompts to report their symptoms, and the app will guide them in managing the identified symptoms. They will also have access to ad hoc symptom reporting (Figure 3) and the ability to track symptom progression (Figure 4). Screenshots of these functionalities are shown in Figures 2-4.

Figure 2. Red-flag symptom reporting.

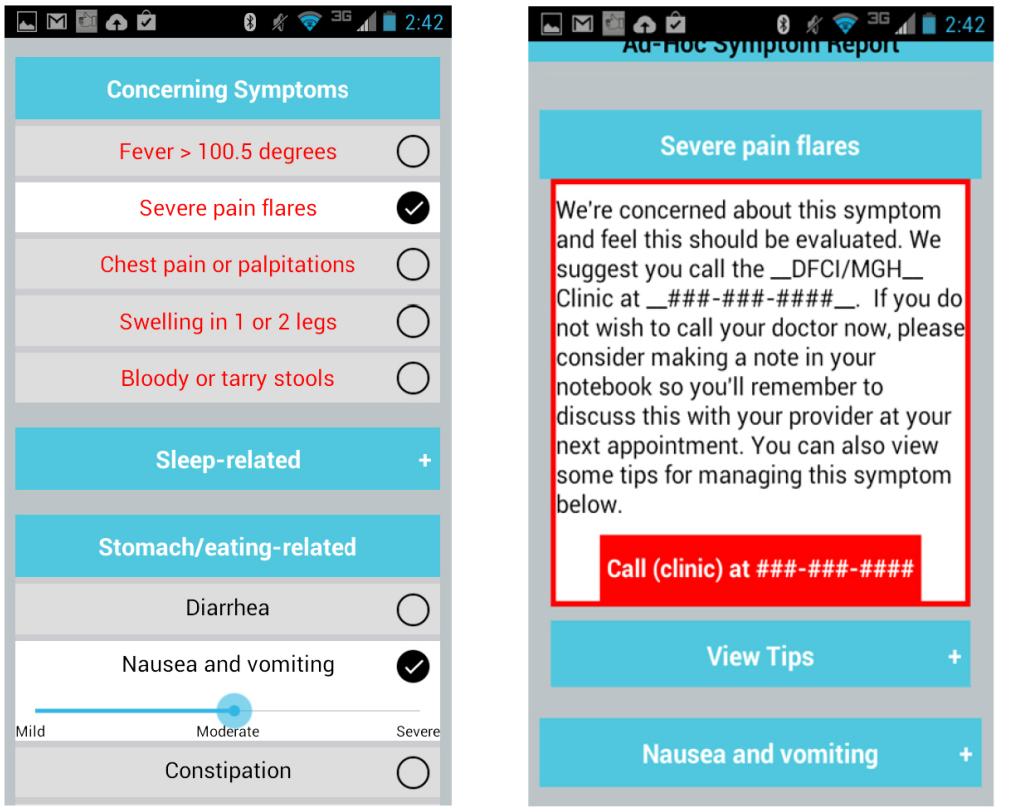


Figure 3. Ad hoc symptom reporting.

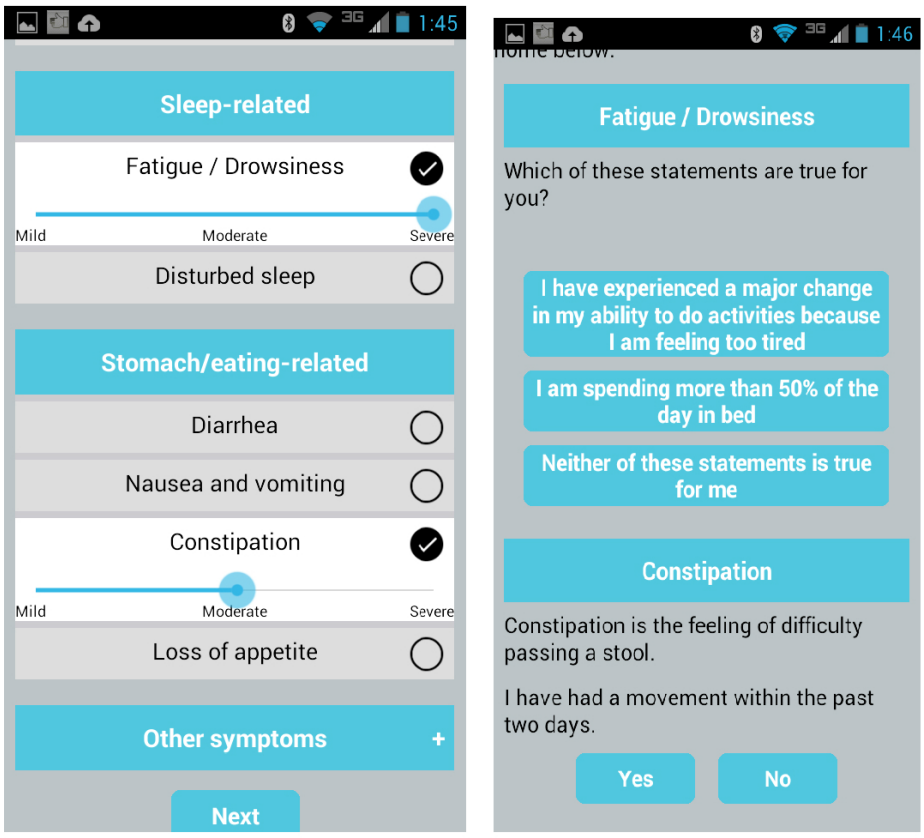
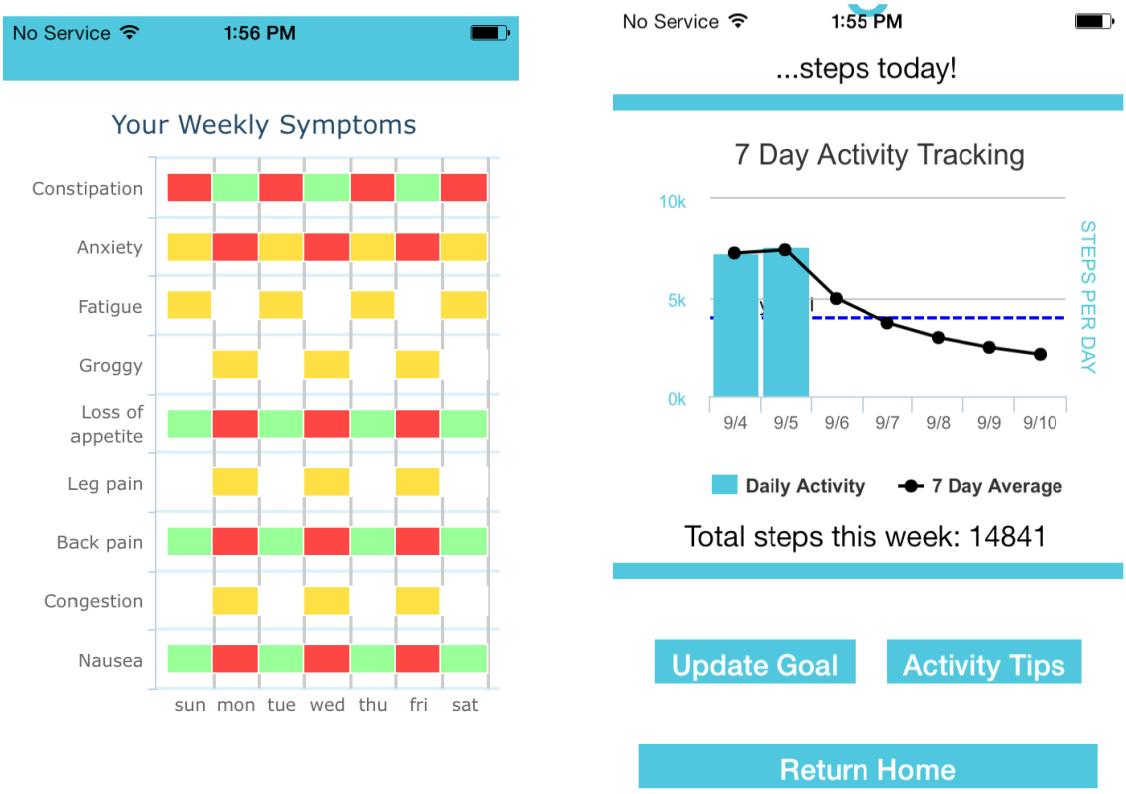


Figure 4. Symptoms and activity tracking.



Symptom and Activity Tracking

My Treatment Plan

Another key function of CORA is the ability of patients to create a treatment plan schedule at enrollment. They will be able enter basic information about their medications and personal preferences like drug names, dosages, scheduled breaks between cycles, food allergies and preferences, etc. They will also be able enter information about other supportive medications like anti-emetics or analgesics. CORA will use all the information entered to help personalize treatment for each participant. For example, a patient on a medication that is required to be taken on an empty stomach will receive a reminder to fast in preparation for the upcoming dose.

Other functionalities include a diary (a notepad integrated in the app allowing patients to log their symptoms or note questions they might have for their care providers at the next hospital visit) and activity tracking (participants will be able to monitor their step counts by using an activity tracker, Fitbit Flex, integrated into the mobile app). We hope that tracking both their activity levels and how they are feeling in general could better help them plan and cope more effectively with the disease.

Treatment Assignments

Intervention Group (CORA + Usual Care)

Subjects assigned to the intervention arm will continue to receive the standard OAMs care at DFCI as usual. In addition to usual care, they will also be able to use the CORA mobile app to manage their medications on their mobile phones for a duration of 3 months.

Control Group

Subjects assigned to the control arm will continue to receive only the standard OAMs care at DFCI as usual.

Randomization and Blinding

A computer program will be used to randomize subjects into the intervention or control arm in a ratio of 1:1 using random permutated blocks to optimize balance in each treatment at any given point in time during the study. Treatment assignment will be concealed in sealed envelopes prepared by a third party not directly involved in the study. Due to the nature of the intervention, it is difficult to blind the subjects to the treatment assignment, but it will be concealed from the investigators and the data analyst. In addition, the study investigators will not be directly involved in the day-to-day running of the study. This will be done by trained research assistants and a project manager who will report regularly to the investigators on the progress of the study.

Outcome Measures

Data Collection Materials

One primary outcome and several secondary outcomes will be assessed in this trial. A number of data collection materials, including validated and study-specific questionnaires, will be used to assess study outcomes at multiple time points over the 3-month period. These tools will be completed in-person under the supervision of the research assistants during the study visits. Among these are the enrollment survey designed by investigators to collect demographic information and subjects'

technology use information, and the Patient-Health Questionnaire (PHQ-8) [18], which will be used as a screener for depression, a potential confounder in this study. If a subject is assessed to have severe depression on this tool, study staff will note this to the file and report immediately to the principal investigator who will duly notify the subject's oncologist by phone. Additionally, such subjects will be encouraged to contact their physicians regarding this screen.

Primary Outcome

The primary outcome measure for this study is medication adherence, defined as the percentage of prescribed doses taken as captured by the MEMS device, which all study participants will receive at enrollment. The MEMS, like other measures of adherence, is not without its drawbacks, but it provides an objective measure of when the pill bottle was opened, which correlates well with the timing of medication self-administration. Currently, there is no gold standard measure of medication adherence, but the MEMS has been used extensively in medication adherence studies [19,20]. Additionally, we will also use a patient self-report tool, the Morisky Medication Adherence Scale (MMAS-8) to measure medication adherence [21]. This is an 8-item self-reported questionnaire used to measure medication adherence. Research has shown that self-reporting of medication adherence captures patient adherence to a reasonably accurate degree. The MMAS has been tested in several different settings and has been shown to demonstrate both concurrent and predictive validity of medication adherence. This survey will be administered in-person by trained research staff at study entry, midpoint, and at the end of the study.

Secondary Outcomes

The following secondary outcomes will be measured at various time points during the study:

- Symptom severity: This will be measured by the MD Anderson Symptom Inventory (MDASI), which is a validated and widely used survey in clinical practice and research [22]. It is used to assess multiple symptoms experienced by cancer patients and how the symptoms interfere with daily living. Its 13 core items include the most frequently occurring symptoms seen in various cancer types and treatment modalities. It will be administered in-person at study entry, midpoint, and end of study. However, because the tool also captures some symptoms that may occur and resolve acutely, we will also assess symptoms in between study visits at weeks 3 and 9. These in-between study assessments will be self-administered via REDCap, a secure, Web-based app for data collection customizable for individual research studies. It is free and complies with all Health Insurance Portability and Accountability Act regulations (HIPAA). It was developed by a multi-institutional consortium initiated at Vanderbilt University. Participants will receive the REDCap links in their in email to complete the surveys online.
- Hospital utilizations: This will be measured at the end of the study by a review of the electronic medical records for emergency department or urgent care clinic visits and in-patient admissions.
- Quality of life: This will be measured longitudinally by the Functional Assessment of Cancer Therapy-General (FACT-G) questionnaire administered in-person at study entry, midpoint, and end of study [23]. Now in its fourth version, this questionnaire is well validated and has been translated into nearly 50 different languages and has been used extensively worldwide. It has four subscales: Physical Well-Being, Social/Family Well-Being, Emotional Well-Being, and Functional Well-Being.
- Cancer-related fatigue: This will be assessed by the Functional Assessment of Chronic Illness Therapy - Fatigue Version 4 (FACIT-F) at study entry, midpoint, and end of study [24]. It is a self-administered 13-item questionnaire validated for use in chronic illness and frequently used with cancer patients.
- Anxiety: The Generalized Anxiety Disorder 7-Item Scale (GAD-7) will be used to assess anxiety [25]. This self-administered 7-item questionnaire is a valid and efficient tool commonly used in both clinical practice and research studies to assess the severity of anxiety.
- Usability and Satisfaction: This is a study-specific questionnaire designed by the authors to collect usability and satisfaction information.

Statistical Analysis Plan

Sample Size Estimation

A sample size of 82 subjects, 41 in each arm (control vs intervention) is sufficient to detect a 10-point difference in mean medication adherence rates (measured by MEMS) between the two groups, assuming equal standard deviation of 16 using a two-tailed *t* test of difference between means with 80% power and a 2-sided alpha of .05. Considering a dropout rate of 20%, the sample size required is 104 (52 subjects per group). Medication adherence rates in the literature vary based on the methods of measurement. The assumptions used in our sample size calculations are based on similar studies, especially those that used the MEMs device as the primary measure of medication adherence [4,6]. A total of 52 participants will be randomly assigned to receive the mobile intervention and will also continue to receive the standard of care for OAMs at the DFCI (intervention group), while the remaining 52 participants will be assigned to the control group that will continue to receive the standard of care at DFCI (usual care group). Participants will be followed up for a total of 3 months.

Statistical Analysis

Data analyses will be done with data analysis and statistical software STATA, version 13, with an alpha of .05 set a priori. Although there are multiple assessment points, all subjects will be followed up for a total of 12 weeks in this 2-parallel group study design. The intention-to-treat approach will be used for all analysis. Descriptive statistics, means (normally distributed) and medians (skewed) continuous data and percentages for categorical variables, will be used to summarize baseline demographic and technology use characteristics by study arm. We will examine for group differences in the primary outcome, percentage of pills taken, by *t* tests or by the non-parametric Mann-Whitney tests if the data are not normally distributed. Logistic regression will be used to identify potential predictors

of adherence. For secondary outcomes, continuous outcomes will be analyzed using *t* test or Mann-Whitney test, and categorical variables will be compared using chi-square tests. Given the longitudinal mode of data collection, a repeated measure analysis of variance will be used to evaluate changes from baseline.

Ethics and Informed Consent

Procedures of our methods have been reviewed and approved by the Dana-Farber/Harvard Cancer Center Institutional Review Board (IRB), and the study will be registered on ClinicalTrials.gov. The app is secure and complies with all HIPPA requirements. Subjects will require a secure passcode to be able to access the app. All mobile phone numbers will be stored in a secure shared-drive available only to IRB-approved study staff. However, if any data breach or adverse effect occurs, the investigator will ensure that they are well documented and reported according to the IRB's requirements, regardless of causality.

Participants will be sent a copy of the consent form in the mail along with the enrollment confirmation letter prior to the enrollment visit. They will be encouraged to review the study procedures and discuss their participation with their families. At the enrollment visit, subjects will be given the necessary time to review the consent form and will be encouraged to ask questions concerning their participation. The research assistant will go through the consent form with the subject to ensure their comprehension of study details. We will make it clear that their participation is completely voluntary and their decision to participate or not participate will not affect their management at DFCI. After the reviewing the consent form and reaffirming their interest in participating in the study, the subject will sign two copies of the consent form, one of which they will take home with them. All enrollment procedures, including the informed consent process, will be done by trained research assistants.

Results

We expect to have results for this study before the end of 2016.

Discussion

Principal Considerations

Our focus on adherence to OAMs is timely because it is estimated that about 25% of cancer therapeutic drugs in development pipelines are oral medications [1]. Also, increased survival and requirement for chronic treatment for some of these cancers like chronic myeloid leukemia will continue to make adherence a prominent issue in the management of patients on OAMS. We hope that CORA can be used in a variety of settings to help patients better manage their medications and symptoms associated with the disease and or medications. Currently, there are only a few studies examining the problem of adherence to OAMs, and many of them are not randomized controlled trials. Although the sample size was small, the study by Simons et al investigating the effect of an intensified multidisciplinary pharmaceutical care program consisting of written and spoken information on adherence to capecitabine in patients with breast or colon cancer found that the mean daily adherence was significantly higher in the intervention group (96.8% vs 87.2%, *P*=.029) and that the intervention also prolonged the chances of patients still being treated with the medication at the end of the follow-up period (48% control group vs 83% in the intervention group; *P*=.019) [20].

Our study is one of the first clinical trials to use a mobile phone app to address the problem of adherence to OAMs. We chose to deploy the app on a smartphone because their adoption is rapidly increasing. Recent national data suggest that more than 56% of Americans now own specifically a smartphone [26]. Another major consideration for deploying this intervention as a mobile app is the additional privacy and security features that can be utilized on smartphones, which helped us to develop a more robust HIPAA-compliant intervention. Users would have to enter a passcode each time they logged on the app.

Limitations

This trial will have limited generalizability of findings given that we are restricting our sample population to patients with renal and prostate cancer. However, CORA was designed for use in patients on any type of OAMs, so we believe that findings from this study could be generalized to other type of cancers. Another threat to generalizability is the fact that we limited study recruitment to one large academic medical center. We believe this should not be of too much concern because the app was designed so that it could easily be adapted for use in other settings.

Conclusion

We have described the trial of a novel solution, deployed on a mobile phone app, for the emerging problem of adherence to OAMs. This innovative approach includes personalizing feedback and management based on patients' own treatment regimen, baseline knowledge, and elucidated barriers to adherence, and holds great promise in improving overall adherence, safety, and clinical outcomes in these patients. We hope that future projects targeted at improving medication adherence in patients on OAMs can build on findings from this project.

Acknowledgments

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Conflicts of Interest

The authors of this article designed CORA but are not responsible for the day-to-day running of the trial.

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Abbreviations

CORA: ChemOtheRapy Assistant
DFCI: Dana Faber Cancer Institute
HIPAA: Health Insurance Portability and Accountability Act regulations
IRB: Institutional Review Board
MEMS: Medication Event Monitoring System
OAMs: oral anti-cancer medications

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Original Paper

A Virtual World Versus Face-to-Face Intervention Format to Promote Diabetes Self-Management Among African American Women: A Pilot Randomized Clinical Trial

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Abstract

Background: Virtual world environments have the potential to increase access to diabetes self-management interventions and may lower cost.

Objective: We tested the feasibility and comparative effectiveness of a virtual world versus a face-to-face diabetes self-management group intervention.

Methods: We recruited African American women with type 2 diabetes to participate in an 8-week diabetes self-management program adapted from Power to Prevent, a behavior-change in-person group program for African Americans with diabetes or pre-diabetes. The program is social cognitive theory–guided, evidence-based, and culturally tailored. Participants were randomized to participate in the program via virtual world (Second Life) or face-to-face, both delivered by a single intervention team. Blinded assessors conducted in-person clinical (HbA1c), behavioral, and psychosocial measurements at baseline and 4-month follow-up. Pre-post differences within and between intervention groups were assessed using t tests and chi-square tests (two-sided and intention-to-treat analyses for all comparisons).

Results: Participants (N=89) were an average of 52 years old (SD 10), 60% had ≤high school, 82% had household incomes <US $30,000, and computer experience was variable. Overall session attendance was similar across the groups (6.8/8 sessions, *P*=.90). Compared to face-to-face, virtual world was slightly superior for total activity, light activity, and inactivity (*P*=.05, *P*=.07, and *P*=.025, respectively). HbA1c reduction was significant within face-to-face (−0.46, *P*=02) but not within virtual world (−0.31, *P*=.19), although there were no significant between group differences in HbA1c (*P*=.52). In both groups, 14% fewer patients had post-intervention HbA1c ≥9% (virtual world *P*=.014; face-to-face *P*=.002), with no significant between group difference (*P*=.493). Compared to virtual world, face-to-face was marginally superior for reducing depression symptoms (*P*=.051). The virtual world intervention costs were US $1117 versus US $931 for face-to-face.

Conclusions: It is feasible to deliver diabetes self-management interventions to inner city African American women via virtual worlds, and outcomes may be comparable to those of face-to-face interventions. Further effectiveness research is warranted.

Trial Registration: ClinicalTrials.gov NCT01340079; http://clinicaltrials.gov/show/NCT01340079 (Archived by WebCite at http://www.webcitation.org/6T2aSvmka).

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KEYWORDS

African Americans; clinical trials; feasibility; health behavior; health disparities; minority health; technology; type 2 diabetes; virtual systems; randomized clinical trial

Introduction

Type 2 diabetes is a complex chronic illness requiring continuing medical care and, ideally, patient adherence to numerous behavioral recommendations for self-management (ie, prescriptions for dietary change, physical activity, weight reduction, blood glucose self-monitoring, smoking cessation, and medication intake) [1] with the goal of achieving glucose control and preventing diabetes complications. Suboptimal control of diabetes places individuals at higher risk for diabetes complications [2].

There are considerable disparities in diabetes risk and outcomes in the population, with African Americans demonstrating among the highest diabetes prevalence and related morbidity and mortality [3,4]. Projected increases in incidence of diabetes may fuel even greater disparities in the future [3]. The traditional medical model involving repeated face-to-face visits over time may represent barriers to diabetes management, especially among underserved populations such as African Americans. Competing family responsibilities, distance to services, transportation difficulties and cost, cost of time away from work and other responsibilities, and difficulties accessing care [5] are among the reasons for limited participation in treatment among patients and may contribute to poor outcomes among African Americans.

With increased penetration rates of Internet use, at 81% in 2013 [6] (up from 71.7% in only 2011) [7], researchers have investigated the impact of delivering behavioral interventions via the Web. The Internet offers alternatives to the challenges typically associated with face-to-face lifestyle interventions through its potential for increased access to specialized behavior change experts, convenience to patients, and potentially lowered costs. However, while online alternatives show promising improvement in health behaviors and glycemic control, effect sizes have been small [8-11]. Limited human interactivity and engagement have been hypothesized as contributors to the small effect sizes [12,13]. In contrast, Virtual world technologies are potentially more suitable environments for supporting diabetes self-management programming. Through the use of three-dimensional (3D) environments that depict real places and avatars that represent people, virtual world environments offer opportunities for interaction, intense engagement, and opportunities for scripted immersive experiences, simulations, role-playing, and constructivist experiences, all important facilitators of active learning [14,15]. The use of virtual world environments continues to increase. There were 1772 million registered virtual world accounts in 2011, with 27 million users registered in Second Life alone [16,17]. The potential of virtual world environments for implementing or supplementing diabetes care interventions has been noted [18], but there is little evidence for the feasibility and potential effectiveness of such an approach [19].

This pilot study examined the feasibility of delivering a group-based diabetes self-management intervention via a virtual world environment (Second Life) and explored the potential effectiveness of the virtual world-based intervention, compared to a traditional face-to-face intervention, on self-management behaviors and glucose levels.

Methods

Design

A randomized clinical trial design was used. A detailed description of the study methods has already been published [20]. The Institutional Review Boards at Boston Medical Center and the University of Massachusetts Medical School approved the trial, and all participants provided written consent prior to participating in the trial.

Participants

Study participants were African American women identified from the medical record data warehouse at Boston Medical Center and affiliated community health centers as having a diagnosis of type 2 diabetes, age ≥18 years, English-speaking, HbA1c>8 at their last outpatient visit (within the previous 12 months), and excluded for medical conditions for which the intervention diet and physical activity would be contraindicated (ie, ulcerative colitis, renal failure, complications following abortion and ectopic and molar pregnancies, angina pectoris, and other forms of unstable ischemic heart disease and other conditions precluding brisk walking). Identified patients were mailed a letter to inform them about the study, to announce a phone call from study staff, and to provide the option to call in or opt out. The staff made up to five calls per patient (on different days and times). Those patients reached were informed about the study (ie, comparison of two formats for delivering a diabetes self-management intervention) and screened for interest and final eligibility (ie, self-reported ability to view a computer screen without difficulty, ability to read, no use of glucocorticoid therapy, no current participation in a weight loss program, and availability for weekly meetings). Fully eligible and interested women were invited to participate and scheduled for an in-person enrollment visit at the Boston Medical Center General Clinical Research Unit. At this visit, participants provided written informed consent and completed baseline assessments.

Randomization

Upon completion of baseline assessment measures, participants were randomized to either the virtual world-based intervention or the face-to-face intervention. Randomization was stratified by age and hemoglobin A1C measured at baseline using a block randomization scheme with a block size of 4, developed by StudyTRAX software (v3.0.0103).

Intervention Conditions

The intervention protocol was similar in both conditions, adapted from the Centers for Disease Control/National Institutes of Health program "Power to Prevent" [21], a widely available social cognitive theory-guided [14], evidence-based, and culturally appropriate behavior-change curriculum designed for delivery to African American groups with diabetes or pre-diabetes via face-to-face group sessions. The intervention sought to enhance diabetes knowledge, optimize attitudes toward diabetes self-management (ie, self-efficacy, outcome expectations), and develop behavioral self-management skills (eg, goal setting, tracking self-management behaviors and glucose levels, problem solving) to facilitate changes in diet, physical activity, blood glucose self-monitoring, and medication adherence. The first session used an individual format followed by eight weekly 90-minute group sessions (group size was 8-9 participants). A single intervention team (a registered dietitian who is a certified diabetes educator, and a nurse practitioner), trained in behavioral counseling and motivational interviewing principles, delivered all sessions in the virtual world environment or face-to-face using the same protocol consisting of a detailed intervention manual and materials (intervention delivery methods are described in greater detail elsewhere) [20]. Intervention fidelity was monitored, and providers were given feedback on behavioral counseling process and content. Participants in both conditions received a two-session computer training and were provided with an Internet-enabled laptop computer upon training completion (Internet access was standardized by providing high-speed 4th generation wireless modems to all participants).

The virtual world-based intervention was delivered in a mock open-air virtual world forum designed and programmed especially for the intervention with appropriate structures and visuals/displays (eg, food exhibits, confidence ruler, a ring of screens, exercise facilities). Participants were asked to log in 30 minutes prior to each session in order to troubleshoot connection or sound problems. A triage system to provide technical support as needed through the session was used. The face-to-face intervention took place in a large conference room at Boston Medical Center. All face-to-face participants received transportation vouchers to facilitate attendance.

Measures and Data Collection

Trained staff, blinded to study condition, conducted assessments at baseline and at 4-month follow-up. Clinical assessments included a non-fasting blood sample for HbA1c assays (specimens were analyzed at the Boston Medical Center laboratories) and measures of blood pressure, height, weight, and waist circumference using standard protocols [20]. At each baseline and follow-up assessment, two telephone-administered unannounced 24-hour recalls assessed diet and physical activity [22,23], blood glucose self-monitoring, and medication adherence. Survey measurements were verbally administered and included measures of depressive symptoms [24], self-efficacy for diabetes management [25], health literacy [26], social support [27], perceived stress [28], quality of life [29], demographics, and other characteristics, including baseline experience with computers and the Internet and post-intervention participant satisfaction. Intervention implementation costs (costs that would be incurred if the intervention were to be implemented outside the context of the research project) were tracked, including staff time, facilities, materials, and set-up (Second Life) for all sessions.

Data Analysis

An intent-to-treat approach was used to compare the virtual world versus face-to-face groups. Feasibility was assessed by comparing the attendance rate by session and the mean number of sessions attended within each arm. Implementation costs were also considered in determining feasibility. Cost estimates were based on expenditures from the trial and excluded the cost of equipment for participants in the virtual world groups. All primary and secondary outcomes were assessed for pre-post differences within each respective arm. The pre-post differences were then compared between the two arms. Differences between continuous variables were assessed using *t* tests. Binomial tests were used for categorical outcomes. Non-parametric tests were applied as appropriate. Analysis of potential mediators was conducted using the taxonomy and recommendations of Zhao [30]. Analyses were conducted in SAS 9.1 and R (version i386 2.153), all comparisons were two-sided and $P<.05$ was considered statistically significant.

We performed bivariate analysis of baseline characteristics to determine whether randomization achieved balance in both treatment groups across all characteristics. The results revealed a statistically significant difference in the proportion of participants with systolic blood pressure greater than 130 mmHg. While multivariate adjustment eliminated the statistical significance of systolic blood pressure at baseline, adjustment did reveal an imbalance in insulin use between the two treatment groups. Using a general linear regression to evaluate the association of insulin use on the pre-post change in HbA1c, we found that it did not have a statistically significant impact and did not affect our assessment of no difference between the virtual world group versus the face-to-face group.

Results

Of the 494 patients who were deemed pre-eligible based on medical records data (age 18 or greater, English-speaking, type 2 diabetes diagnosis, last HbA1c>8 within previous 12 months), it was not possible to determine the eligibility of 321 patients for reasons listed in Figure 1. Of the 174 (35%) who were reached for telephone screening, 62 (36%) were ineligible and 112 (64%) were eligible. From these 112 patients with known eligibility, 89 (79%) were enrolled and randomized, 46 of them to the virtual world intervention and 43 to the face-to-face intervention.

Table 1 summarizes demographic and baseline characteristics of participants in the trial: average age was 52 years (SD 10) and 90% of participants were over the age of 40; 60% had a high school education or lower; 82% reported a household income of US $30,000 or less; and experience with computers was variable. The single statistically significant difference between the virtual world and face-to-face groups at baseline was the proportion of participants with systolic blood pressure

greater than 130 mmHg. A greater proportion of participants in the face-to-face group had elevated systolic pressure compared to those in the virtual world group (18% vs 10%, *P*=.04).

Figure 1. Flowchart of participant screening, recruitment, randomization, and retention.

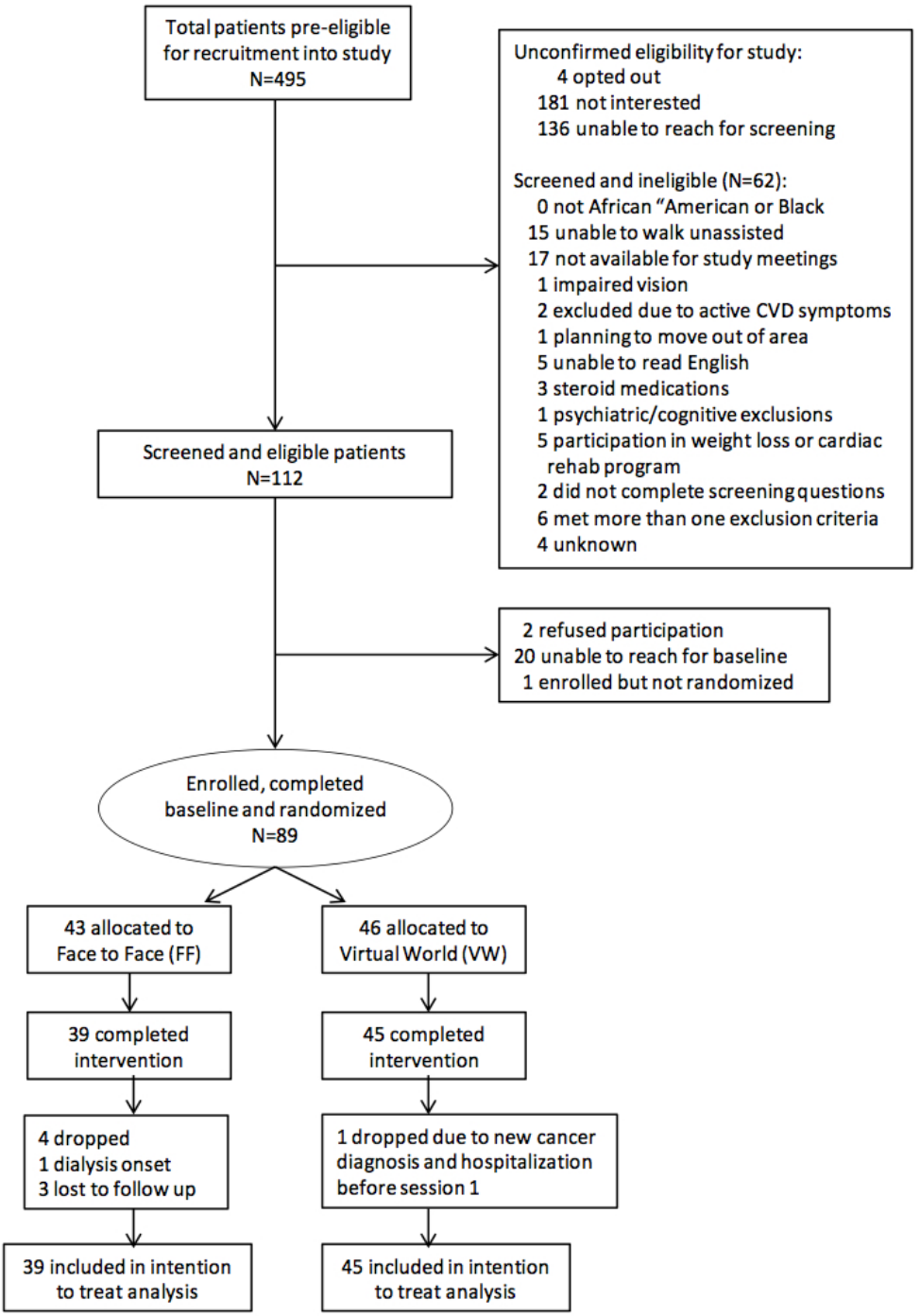

Table 1. Sample characteristics at baseline.

	All subjects (N=89)	virtual world (n=46)	Face-to-face (n=43)	*P*
Demographic characteristics				
Age in years, mean (SD), IQR	52 (10), 49-58	53 (10), 49-59	52 (11), 48-57	.77
18-40, n (%)	9 (10.1)	6 (13.0)	3 (7.0)	.49[a]
>40, n (%)	80 (89.9)	40 (87.0)	40 (93.0)	
Marital status, n (%)				
Single (never married)	45 (50.6)	27 (58.7)	18 (41.9)	
Married or living with partner	19 (21.4)	9 (19.6)	10 (23.3)	
Separated, divorced, or widowed	25 (28.0)	10 (21.7)	15 (34.8)	
Education in years, mean (SD), IQR	13.1 (2.2), 12-16	13.3 (2.3), 12-16	12.8 (2.0), 12-14	.30
<High school, n (%)	16 (18.0)	7 (15.2)	9 (20.9)	.44
High school graduate, n (%),	37 (41.6)	17 (37.0)	20 (46.5)	
Vocational/Assoc degree, n (%)	13 (14.6)	9 (19.6)	4 (9.3)	
≥College, n (%)	23 (25.8)	13 (28.3)	10 (23.3)	
Work status, n (%)				
Working full or part-time	32 (36.0)	13 (28.3)	19 (44.2)	.12
Not working	57 (64.0)	33 (71.7)	24 (55.8)	
Household income, n (%)				
≤$10,000	28 (31.5)	16 (34.8)	12 (27.9)	.82
$10,000-$30,000	45 (50.5)	23 (50.0)	22 (51.2	
≥$30,000	10 (11.2)	6 (13.0)	4 (9.3)	
Declined	6 (6.7)	1 (2.2)	5 (11.6)	
Insurance, n (%)				
Public/no insurance	68 (76.4)	37 (80.4)	31 (72.0)	.35
Private	21 (23.6)	9 (19.6)	12 (27.9)	
Health literacy (confidence filling out medical forms by herself), n (%)				
Extremely	61 (72.6)	37 (82.2)	24 (61.5)	.19[a]
Quite a bit	6 (7.1)	1 (2.2)	5 (12.8)	
Somewhat	10 (11.9)	4 (8.9)	6 (15.4)	
A little	5 (5.9)	2 (4.4)	3 (7.7)	
Not at all	2 (2.4)	1 (2.2)	1 (2.6)	
Computer experience				
Hrs/wk using a computer, mean (SD)	13 (17)	11 (12)	15 (21)	.39
Home Internet access, n (%)				
Yes	61 (68.54)	34 (73.91)	27 (62.79)	.26
No	28 (31.46)	12 (26.09)	16 (37.21)	
Able to start and shut down computer on her own, n (%)				
Not at all	5 (5.62)	3 (6.52)	2 (4.65)	.59[a]
With a lot of help	3 (3.37)	1 (2.17)	2 (4.65)	
With a little bit of help	18 (20.22)	7 (15.22)	11 (25.58)	
Without help	63 (70.79)	35 (76.09)	28 (65.12)	
Able to create and send email on her own, n (%)				

	All subjects (N=89)	virtual world (n=46)	Face-to-face (n=43)	*P*
Not at all	13 (14.61)	5 (10.87)	8 (18.60)	.18[a]
With a lot of help	8 (8.99)	3 (6.52)	5 (11.63)	
With a little bit of help	20 (22.47)	8 (17.39)	12 (27.91)	
Without help	48 (53.93)	30 (65.22)	18 (41.86)	
Able to go online on the Internet on her own, n (%)				
Not at all	9 (10.11)	2 (4.35)	7 (16.28)	.16
With a lot of help	8 (8.99)	5 (10.87)	3 (6.98)	
With a little bit of help	11 (12.36)	4 (8.70)	7 (16.28)	
Without help	61 (68.54)	35 (76.09)	26 (60.47)	
Use Internet to search for health information, n (%)				
Never	33 (37.08)	13 (28.26)	20 (46.51)	.19[a]
Rarely	8 (8.99)	4 (8.70)	4 (9.31)	
Occasionally	30 (33.71)	20 (43.48)	10 (23.26)	
Often	18 (20.22)	9 (19.57)	9 (20.93)	
Use Second Life (yes)	3 (3.37)	0	3 (6.98)	.11
Health characteristics, n (%)				
HbA1c				
<7%	7 (7.9)	4 (8.7)	3 (7.0)	1.0
7-7.9%	14 (15.7)	7 (15.2)	7 (16.3)	
8-8.9%	13 (14.6)	7 (15.2)	6 (13.9)	
≥9.0%	55 (61.8)	28 (60.9)	27 (62.8)	
Insulin usage	44 (49.4)	21 (45.7)	23 (53.5)	.46
BMI (kg/m ^{2}), n (%)				
Normal (<25)	4 (4.5)	2 (4.4)	2 (4.7)	.82
Overweight (25-29.9)	18 (20.2)	8 (17.4)	10 (23.3)	
Obese I (30-34.9)	27 (30.3)	13 (28.3)	14 (32.6)	
Obese II (35-39.9)	21 (23.6)	11 (23.9)	10 (23.3)	
Obese III (≥40)	19 (21.3)	12 (26.1)	7 (16.3)	
Waist circumference >35 in, n (%)	80 (89.9)	41 (89.1)	39 (90.7)	1.0[a]
Systolic blood pressure >130, n (%)	28 (31.5)	10 (21.7)	18 (41.9)	.04
Diastolic blood pressure >80, n (%)	48 (53.9)	22 (47.8)	26 (60.5)	.23
Total cholesterol >200, n (%)	29 (32.6)	13 (28.3)	16 (37.2)	.37

[a]*P* value derived from Freeman-Halton extension of Fisher's exact test.

Feasibility of the Virtual World-Based Intervention

Overall session attendance was similar across the two interventions, with an average 6.8 sessions (SD 1.8) among WV participants and 6.8 (SD 1.7) among face-to-face participants (*P*=.9). However, a significant difference was observed in the rate of completion of Session 1. Compared to face-to-face participants, fewer virtual world participants completed this session (78%, 36/46 vs 95%, 41/43, *P*=.02, respectively). There was also a slight although non-significant difference in the proportion of participants completing sessions 1-3 in the virtual world group (63%, 29/46) versus the face-to-face group (77%, 33/43) (*P*=.16).

Overall participant retention rate was 94% for clinical and psychosocial assessments, and 93% for telephone-based assessments, and attrition was lower in the virtual world group (1 participant, or 2%) compared to the face-to-face group (4 participants, or 9%) (*P*=.19). The one drop-out in the virtual world group was due to a new cancer diagnosis and unexpected hospitalization and occurred prior to session 1. Reasons for drop-out in the face-to-face group included dialysis onset and loss to follow-up (2 of these 4 participants attended Session 1).

Health Outcomes

Results from intention-to-treat analyses are shown in Table 2. There were improvements associated with both the virtual world and the face-to-face intervention conditions. Analysis of change from baseline to 4-month follow-up within the groups revealed a non-statistically significant 3.2% reduction in HbA1c in the virtual world group (*P*=.186) and a significant 4.9% reduction in HbA1c in the face-to-face group (*P*=.019). However, no significant differences between the groups were detected (*P*=.52). There was also a statistically significant within group decrease in the percentage of participants with HbA1c ≥9% in both groups, with 14% fewer participants in each group having a HbA1c value above 9.0% (*P*=.014 and *P*=.002, for virtual world and face-to-face groups, respectively), with no significant differences between the groups (*P*=.493). No significant within or between-group changes were observed in measures of blood pressure, total cholesterol, waist circumference, and Body Mass Index (BMI).

Behavioral Outcomes

Participants in the virtual world group experienced an 18.4% within-group increase in total physical activity and a significant 8% decrease in inactivity (*P*=.10 and *P*=.04, respectively), whereas participants in the face-to-face group experienced a 22.5% reduction in their total physical activity. There was a marginally significant between difference in total physical activity, with marginally superior effects for the virtual world compared to face-to-face group on total activity, light activity, and inactivity (*P*=.05, *P*=.07, and *P*=.025, respectively). The proportion of participants not adhering to blood glucose self-monitoring dropped by half in both groups (*P*=.001 and *P*=.002 for virtual world and face-to-face, respectively), with no significant between-group differences for this outcome. No significant within or between-group differences were observed for dietary outcomes of interest (ie, total calories, percent calories of saturated fat, fiber or dietary quality as measured by the Alternate Healthy Eating Index). Medication adherence decreased in the face-to-face group with 8.6% fewer participants reporting that they adhered to all medications as prescribed (*P*=.035), whereas there was a 1.2% increase in the virtual world group, although no differences between the groups with regards to change in self-reported medication adherence (*P*=.298).

Psychosocial Outcomes

Depression symptom scores and mental health functioning (as measured by the Short-Form survey [SF-12]) were marginally improved in the face-to-face condition only (*P*=.053 and *P*=.062, respectively), and there were between-group differences for depression symptom score pre-post change (*P*=.051). Improvements in self-efficacy for diabetes self-management were observed in both groups (*P*<.001), and there were no differences in self-efficacy improvements between the groups (*P*=.268). No within or between-group differences were observed for perceived stress or social support.

Mediation Analysis

We evaluated the potential mediation effects of select behavioral and psychosocial outcomes and found insufficient evidence of a mediation effect on HbA1c levels for changes in total calories consumed, total calories from saturated fat, total dietary fiber, alternate healthy eating index, diabetes self efficacy scores, inactivity levels, activity levels (household, light, and moderate activity), medication adherence post intervention, and blood glucose self-monitoring (data not shown).

Intervention Costs

The per-participant cost of implementing the virtual world intervention was US $186.39 greater compared to cost of implementing the face-to-face intervention (US $1117 vs $931 for virtual world vs face-to-face, respectively). Expenses associated with health care personnel (eg, diabetes nurse educator, dietitian, administrative staff) and educational materials were the same between the two groups. However, the virtual world group required additional technical personnel who trained and provided technical assistance to participants and the intervention team during each session, contributing to 13% of the total cost per participant in that group.

Participant Satisfaction

At the follow-up assessment, 97% of face-to-face participants agreed/strongly agreed with the statement “If I had a choice, I would attend diabetes sessions face to face rather than on a computer”, while 80% of virtual world participants agreed/strongly agreed with the statement “If I had a choice, I would attend diabetes sessions on Second Life rather than face-to-face at BMC or my health center” (*P*=.490). However, there were no differences between the groups with regard to whether they would recommend their program to other people; 100% of virtual world participants agreed/ strongly agreed with the statement “I would recommend other people to attend diabetes education sessions given in Second Life,” and 97% of face-to-face participants agreed/ strongly agreed with the statement “I would recommend other people to attend diabetes education sessions given face to face at BMC or a health center” (*P*=1.0).

Table 2. Within group and between group comparisons for the virtual world and face-to-face intervention conditions (*P* values for continuous variables derived from Student's *t* test of pre-post differences, unless otherwise noted).

Variable	Face-to-face					Virtual world					
	Baseline (n=43)	Post (n=39)	Baseline/ follow-up difference	% change	Within group, *P*	Baseline (n=46)	Post (n=45)	Baseline/ follow-up difference	% change	Within group, *P*	Between group, *P*
Clinical outcomes, mean (SD)											
HbA1c	9.4 (2)	8.9 (2)	-0.46	-4.9	.019	9.6 (2)	9.3 (2)	-0.31	-3.2	.186	.519
HbA1c<9[a,b], %	37.2	51.3			.002	39.1	53.3			.014	.493
Systolic BP, mmHg	126.0 (15)	126.0 (17)	0.05	0.04	.808	120.5 (13)	122.3 (16)	1.81	1.5	.233	.609
Diastolic BP, mmHg	80.4 (11)	78.7 (9)	-1.64	-2.0	.733	79.4 (9)	80.1 (10)	0.72	0.9	.600	.675
Cholesterol	194.6 (42)	191.1 (40)	-3.50	-1.8	.186	187.8 (49)	186.9 (45)	-0.90	-0.5	.971	.412
BMI	34.4 (8)	33.4 (6)	-1.00	-2.9	.912	36.4 (8)	36.1 (8)	-0.30	-0.8	.134	.200
Waist circumference (in)	110.0 (17)	107.2 (15)	-2.80	-2.5	.631	113.1 (17)	112.1 (16)	-1.03	-0.9	.415	.837
Behavioral variables (weekday)											
Self-reported diabetes medication adherence, %											
All prescribed diabetes medications[a,b]	88.1	79.5			.035	86.9	88.1			.488	.2984
Dietary intake, weekday averages, mean (SD)											
Total calories, kcal	1377.0 (512)	1181.3 (443)	-195.67	-14.2	.023	1220.7 (518)	1136.0 (492)	-84.70	-6.9	.257	.255
% calories from SFA	11.0 (4)	9.8 (4)	-1.17	-10.6	.162	10.7 (4)	9.9 (3)	-0.78	-7.3	.434	.544
Fiber	11.4 (6)	13.1 (8)	1.63	14.2	.163	13.0 (7)	13.6 (7)	0.59	4.5	.627	.496
Alternate Healthy Eating index	27.0 (9)	29.8 (9)	2.86	10.6	.121	29.4 (10)	30.6 (10)	1.16	3.9	.548	.469
Physical activity (PA), weekday averages, mean (SD)											
Total PA (MET-hr)	40.9 (32)	31.7 (29)	-9.20	-22.5	.196	36.5 (27)	43.2 (31)	6.70	18.4	.113	.050
Total inactivity (MET-hr)	63.0 (13)	65.2 (13)	2.24	3.5	.269	65.0 (12)	60.0 (15)	-5.00	-7.7	.040	.025
Household activity (MET-hr)	18.0 (16)	11.8 (11)	-6.20	-34.4	.013	16.0 (11)	14.0 (11)	-2.00	-12.5	.318	.140
Light activity (MET-hr)	36.0 (24)	28.6 (29)	-7.40	-20.6	.311	33.0 (27)	39.2 (32)	6.24	18.9	.101	.071
Moderate activity (MET-hr)	16.0 (20)	15.9 (28)	-0.15	-0.9	.650	16.0 (26)	19.5 (29)	3.50	21.9	.363	.472
Median	8.0	8.3				6.0	8.3				
Blood glucose self-monitoring [a], %											
No monitoring	35.0	15.0			.002	24.0	12.0			<.001	.895
Psychosocial variables, mean (SD)											
Depressive symptoms (CES-D)	22.6 (9)	20.6 (8)	-2.01	-8.9	.053	19.8 (9)	20.5 (10)	0.71	3.6	.441	.051

Variable	Face-to-face					Virtual world					
	Baseline (n=43)	Post (n=39)	Baseline/ follow-up difference	% change	Within group, *P*	Baseline (n=46)	Post (n=45)	Baseline/ follow-up difference	% change	Within group, *P*	Between group, *P*
Perceived stress (PSS)	14.2 (7)	13.9 (8)	-0.28	-2.0	.263	14.2 (8)	15.1 (7)	0.87	6.1	.336	.139
Physical functioning (SF-12 PCS)	41.7 (9)	43.9 (11)	2.18	5.2	.247	42.4 (10)	42.3 (11)	-0.08	-0.2	.813	.322
Mental health functioning (SF-12 MCS)	47 (10)	50 (11)	3.74	8.0	.062	49 (11)	50.3 (12)	1.32	2.7	.385	.293
Overall quality of life (SF-12 total score)	88.5 (13)	94 (14)	5.88	6.6	.022	91.3 (16)	92.6 (14)	1.24	1.4	.599	.113
Social support	68.0 (20)	72 (20)	3.50	5.1	.261	69.5 (27)	67.7 (28)	-1.79	-2.6	.602	.256
Diabetes self-efficacy	34.6 (7)	40 (7)	5.90	17.1	<.001	36.0 (9)	40.6 (7)	4.58	12.7	<.001	.268

[a]Within group differences determined by Fisher's Exact test.

[b]Between group differences determined by Breslow-Day Test for Homogeneity.

Figure 2 contains pictures of virtual world intervention sessions and edited clips of the sessions (see also Multimedia Appendix 1).

Figure 2. Pictures of the Virtual World participants engaged in various intervention sessions.

Discussion

Principal Findings

To our knowledge, this pilot randomized controlled trial (RCT) is the first study to compare the feasibility and potential comparative effectiveness of a virtual world-based versus an face-to-face-based diabetes self-management intervention. A previous publication reported on the feasibility of conducting individual virtual world-based visits with participants with diabetes, but the intervention did not include lifestyle

modification, the study did not include a comparison condition, and metabolic outcomes were not reported [19]. Our study showed that it is feasible to deliver a virtual world group-based behavioral intervention originally designed for face-to-face delivery, to improve diabetes self-management among inner-city African American women. All participants who began the virtual world group completed the study, whereas 4 participants in the face-to-face condition did not.

Study findings show that the virtual world technology has tremendous potential for intervening and improving glucose control and diabetes self-management behaviors. Reductions in glucose levels were similar across both groups and were comparable to those of other group interventions [31]. Furthermore, HbA1c reductions were particularly significant among participants with the highest baseline glucose control (HbA1c≥9%).

Our study showed a marginal superiority of the virtual world intervention, compared to the face-to-face intervention, on physical activity (increase in total and light physical activity, and reduction in inactivity). A greater effect of the virtual world over the face-to-face intervention format on physical activity was also reported by Johnston et al [13]. In that study, the authors attributed this effect to the virtual world environment facilitating opportunities for the individual to initiate and practice healthy behaviors through an avatar with whom they identify. A study by Napolitano et al [32] that explored the usability of avatars for modeling weight loss behaviors provided additional support for the potential of virtual worlds for influencing diet and exercise behaviors. Two additional studies reported promising results from virtual world-based interventions for smoking cessation among rural teens [33,34]. Known as the Proteus effect, the practice of a new behavior by one's avatar may influence the individual's behavior in the real world [35].

The virtual world and face-to-face interventions were both comparable in terms of fostering blood glucose self-monitoring and enhancing diabetes management self-efficacy, and the face-to-face intervention was marginally superior compared to virtual world with regards to reducing depression symptoms. Social support interventions have improved depressive symptoms among people with diabetes, and it is possible that the face-to-face interaction influences participants in a different manner compared to virtual world interactions, potentially facilitating greater or a different type of social support [36].

Strengths and Limitations

There were technical challenges in the virtual world intervention that affected completion of the first session. Additional technical difficulties occurred with decreasing frequency over the course of the intervention. The technical support during the virtual world sessions was necessary to assure that each participant was able to navigate, hear, and interact in the virtual world. The two most commonly encountered technical challenges were strength of Internet connection and sound problems (could not hear or could not speak). virtual worlds demand significant bandwidth as they process enormous amounts of data to render the 3D spaces, physical interactions, and sound that characterize these environments, and our chosen Internet service, provided via wireless modem, was unreliable in the participants' neighborhoods (inadequate cell tower coverage). We devised a triage system troubleshooting these problems (re-positioning in the home, and checks for headphone, laptop, and Second Life preferences options). Undoubtedly, virtual world platforms are rapidly improving and becoming more accessible and technically efficient.

Despite these challenges, participants seemed to be equally satisfied and engaged in both intervention groups. Consistent with the high level of satisfaction reported by participants in both groups, attrition was remarkably low in this hard-to-reach group of African American women. A previous study of a virtual world-based versus face-to-face-based intervention that targeted weight loss in a non-minority, educated and more affluent group reported a 13% drop-out rate in both groups, with 5 virtual world participants reportedly dropping out within the first 2 weeks of the program for reasons associated with technical difficulties [13].

The per-participant cost of the virtual world intervention was 13% higher than that of the face-to-face intervention, with the excess cost related primarily to the need for technical support staff to train and support our participants, as the study participants had variable levels of computer experience. This study is unable to answer the question of whether the increased cost of the virtual world intervention outweighs its potential benefit; however, it does provide evidence of feasibility and preliminary evidence of effectiveness for future larger RCTs that can answer such questions. It is important to note that the cost of virtual world interventions should be expected to decrease over time with improved technologies and Internet access for the wider population.

A particular strength of the study was the use of the virtual world format with a socioeconomically disadvantaged sample of African American women. African Americans constitute a high risk group with a high prevalence of diabetes, diabetes complications, and mortality [3,4]. The sample was middle-aged with most women having a high school education or lower, low household income, and variable computer experience. Furthermore, the sample consisted of 79% of participants who were reached and for whom eligibility was known, supporting the representativeness of the sample and potential generalizability of the study findings. Prior studies of Internet-based interventions have included primarily young individuals. While it has been hypothesized that demographic differences (including age, ethnicity, income, and culture) could impact the effect of interventions [37], our study found that the virtual world intervention was feasible and had benefits for our African American sample. The generalizability of Internet-based interventions also has been questioned based on potential selection bias by which individuals with low computer or Internet literacy may refrain from participation, may be excluded by the study's eligibility criteria, or may more easily drop out from these interventions. For example, the only prior pilot study comparing a virtual world-based versus face-to-face-based interventions for weight loss [13] recruited virtual world participants via print and online media and excluded participants who had no access to an Internet-connected computer (73% of participants held college or advanced degrees and had incomes

above $75,000). In contrast, our study systematically recruited patients from community health centers and a large safety net hospital using electronic databases, and minimized exclusion criteria in an effort to provide accurate data on the feasibility and outcomes of the virtual world intervention for inner-city African American women. The fact that the sample had variable computer experience and most participants had no prior exposure to virtual world environments provides further support for the potential generalizability of virtual world-based behavioral interventions.

Additional study strengths include the parallel content and structure of the virtual world and the face-to-face interventions, and their delivery by a single provider team, with only the intervention format being different across groups, and the tracking of cost data for both groups. While the possibility of reduced care cost has been an argument for Web-based interventions, very few studies have compared the costs associated with the implementation of virtual world versus face-to-face.

Conclusions

Future research is needed to test the comparative effectiveness of the virtual world and face-to-face interventions in larger, appropriately powered trials and with a longer follow-up. Future studies should also investigate characteristics of individuals who do best with each one of the two approaches. For example, it has been suggested that men may have a stronger experience of "presence" (ie, perceived realism, sense of being present) when being immersed in virtual world environments [38,39]. Furthermore, there is currently little understanding of potential mechanisms that facilitate health behavior change and adherence in virtual world environments. Future studies need to examine virtual world environment and avatar factors that facilitate health behavior change, including the degree to which one's experience in the virtual world influences one's behavior in the real world (Proteus effect) [35]. In 2010, the National Heart Lung and Blood Institute (NHLBI) convened a workshop entitled "Virtual Reality Technologies for Research and Education in Obesity and Diabetes", which included behavioral and health researchers, technology experts, and representatives of the other National Institutes of Health institutes (National Cancer Institute, National Institute of Child Health and Human Development, National Institute of Diabetes and Digestive and Kidney Diseases, the NIH Office of Behavioral and Social Sciences Research, and the NIH Office of Research on Women's Health). A report [40] from this workshop identified a number of research priorities, including the impact of using virtual reality technologies for fostering health-related behaviors and for extending the availability and capacity of health care providers (ie, "extended classrooms for diabetes education"). This study addressed both priorities.

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Conflicts of Interest

None declared.

Multimedia Appendix 1

Link to video of Virtual World intervention sessions.

[PDF File (Adobe PDF File), 15KB - resprot_v3i4e54_app1.pdf]

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Abbreviations

BMI: body mass index
BP: blood pressure
CES-D: Center for Epidemiologic Studies Depression Scale
MET: metabolic equivalent of task
PA: physical activity
PCS and MCS: Physical and Mental Health Composite Scores
PSS: Perceived Stress Scale
SF-12: short-form survey
SFA: saturated fatty acids

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Original Paper

Promoting Physical Activity in Low-Active Adolescents via Facebook: A Pilot Randomized Controlled Trial to Test Feasibility

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Abstract

Background: The World Wide Web is an effective method for delivering health behavior programs, yet major limitations remain (eg, cost of development, time and resource requirements, limited interactivity). Social media, however, has the potential to deliver highly customizable and socially interactive behavioral interventions with fewer constraints. Thus, the evaluation of social media as a means to influence health behaviors is warranted.

Objective: The objective of this trial was to examine and demonstrate the feasibility of using an established social networking platform (ie, Facebook) to deliver an 8 week physical activity intervention to a sample of low-active adolescents (N=21; estimated marginal mean age 13.48 years).

Methods: Participants were randomized to either an experimental (ie, Behavioral) or attentional control (ie, Informational) condition. Both conditions received access to a restricted-access, study-specific Facebook group where the group's administrator made two daily wall posts containing youth-based physical activity information and resources. Primary outcomes included physical activity as assessed by accelerometry and self-report. Interactions and main effects were examined, as well as mean differences in effect sizes.

Results: Analyses revealed significant improvements over time on subjectively reported weekly leisure-time physical activity ($F_{1,18}$=8.426, P=.009, η2 = .319). However, there was no interaction between time and condition ($F_{1,18}$=0.002, P=.968, η2 = .000). There were no significant time or interaction effects among the objectively measured physical activity variables. Examination of effect sizes revealed moderate-to-large changes in physical activity outcomes.

Conclusions: Results provide initial support for the feasibility of delivery of a physical activity intervention to low-active adolescents via social media. Whether by employing behavioral interventions via social media can result in statistically meaningful changes in health-related behaviors and outcomes remains to be determined.

Trial Registration: ClinicalTrials.gov NCT01870323; http://clinicaltrials.gov/show/NCT01870323 (Archived by WebCite at http://www.webcitation.org/6SUTmSeZZ).

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KEYWORDS

social media; physical activity; adolescents; behavior change; randomized controlled trial; feasibility

Introduction

Physical Activity and Adolescents

Physical activity levels among adolescents are at all time lows [1-3], with less than one in five American adolescents meeting the recommended guidelines of accumulating at least 60 minutes of moderate-to-vigorous physical activity on a daily basis [4,5]. It is well established that increased involvement in various forms of physical activity (eg, exercise, sport, play, leisure, transportation) is associated with an array of health-related benefits including, but not limited to, physical health (eg, fat loss and musculoskeletal health), mental health (eg, reduced anxiety and improved self-esteem), cognitive health (eg, increased academic achievement), and behavioral health (eg, favorable adolescent risk profiles) [6-13].

Traditionally, physical activity interventions for youth have often used in-person, center-based modes of delivery [14,15]. Although these face-to-face interventions have been identified as the "gold standard" of behavioral therapy, they are limited in terms of reach and accessibility. Fortunately, the relatively recent introduction of the Internet into clinical practice has created several opportunities for innovative behavioral interventions [16,17]. Researchers have become increasingly more interested in using the Internet as a mode of delivery for physical activity programs [15,18], and a number of reviews attest to the feasibility and efficacy of Internet-delivered interventions for changing health behavior outcomes [19-21].

More recently, advances in Web-based programing have led to interactive communication technologies, commonly referred to as *social media*. These sites provide users with the ability to make virtual connections and interact with one another via user-generated content [22-24]. Due to the ease of use, accessibility, minimal cost, limited maintenance (on the user's end), and various interactive communication features, these relatively new technologies have quickly gained universal acceptance [25]. As a result, researchers have begun to examine the potential of social media as a means for increasing education and social support for individual behavior change. Emerging evidence suggests that physical activity interventions using social media can be effective at influencing health and promoting behavior change [21,25-29]. Social media sites provide users with the ability for continuous self-monitoring, real-time feedback, and information exchange, all of which are conducive to behavior change [17]. Thus, further evaluation of social media as a means to influence health behaviors is warranted [30-33].

Social Networking Sites and Adolescents

Social networking sites are one of the most popular forms of social media [27,34], especially among adolescents [32,35,36], and are becoming an alternative to email as a means for instant communication between friends [37]. Facebook, in particular, is the most widely used social networking site by adolescents or any other demographic, reaching roughly two thirds of the Internet population [38]. Adolescents, 13-17 years old, make up nearly 21% of the Facebook population [35]. Sites such as Facebook have become a prominent source of information and guidance during adolescence. For example, 57% of adolescents look to their social networking sites for advice, making them 63% more likely to do so than the typical social networker [34]. These vital communication hubs have the information and tools necessary for developing and managing healthy lifestyles, with roughly one third of online adolescents using the Internet for health-, diet-, or fitness-related information [36]. Thus, social networking sites may hold the potential to aid in the promotion of health and encourage changes in behavior [21,31-33].

The Present Study

The *Social Media and Activity Research in Teens* (*SMART*) Trial is a social media-based intervention specifically designed to influence the physical activity behaviors of adolescents. The purpose of this innovative 8 week program was to test the feasibility of delivering a physical activity intervention to low-active adolescents over an established social media platform (ie, Facebook). Furthermore, we were interested in examining the differences in behavioral outcomes between two social media-based conditions (ie, an experimental group which received behavioral training modules vs an attentional control group). It was hypothesized that exposure to and participation in this particular intervention would provide initial support for the potential of using social media to promote positive changes in physical activity behaviors among a sample of low-active adolescents. Additionally, exposure to video-based behavioral training modules should lead to greater improvements in physical activity participation than simple exposure to physical activity-related information and resources.

Methods

Study Design and Participants

An 8 week randomized controlled pilot trial was designed to increase lifestyle physical activity in adolescents (NCT01870323). Participants were recruited from Champaign County, Illinois. Individuals who met inclusionary criteria were randomized (matched by age and sex) to either the intervention (ie, sharing physical activity-related content via Facebook along with weekly behavioral modules; *Behavioral Condition*) or attentional control group (ie, sharing physical activity-related content via Facebook alone; *Informational Condition*).

Recruitment

The Internet (ie, laboratory website recruitment page, emails of former parental research participants, campus-wide listservs aimed at faculty and staff, and Facebook pages of local organizations) and targeted mailings, via the United States Postal Service, were used for recruitment purposes. Given that this trial relied heavily on parental involvement, recruitment efforts were targeted at parents or legal guardians of children between the ages of 13 and 15 years old. Advertisements for recruitment included basic information about the study, along with contact information (ie, study-specific email and website) for interested parties. To be considered for participation, a parent or legal guardian had to accompany all eligible adolescent participants. Parental guardians were screened via telephone to determine the physical activity levels of their children. Those meeting or exceeding federal guidelines were excluded. Additionally, participants were required to be English-speaking and have

access to the Internet at their place of residence via a personal or family-dedicated tablet, laptop, or desktop computer. Individuals who only had mobile access to the Internet were excluded, to ensure similar user experience (although accessing the Facebook group via a mobile application was permitted). Finally, participants also had to have an active Facebook account or be willing to create one prior to enrollment.

SMART Facebook Group

Access to a single, study-specific Facebook group (ie, the *SMART* Group) was restricted to all study participants and at least one of their legal guardians. The purpose of this group was to create a social, interactive community that revolved around the topic of physical activity for youth. Physical activity-related information and resources from around the Web were provided daily by the group's administrator. Considering the varying characteristics and preferences of the adolescent sample, an assortment of group wall posts (ie, readily available information shared in a virtual community) were made, all of which were categorized within one of the following seven categories: (1) physical activity-related websites; (2) infographics; (3) public service announcements (PSA); (4) technology and applications; (5) local parks and facilities; (6) motivational quotations; and (7) miscellaneous topical posts. The *SMART* Group received two wall posts per day (ie, once in the morning and again in the evening), resulting in 14 total wall posts per week with equal distribution of the predefined content categories (see Table 1). In addition to these daily posts, photo albums containing physical activity campaign advertisements/posters (eg, The President's Challenge) were uploaded and shared once per week, resulting in eight additional posts. In all, 120 posts were made over the course of the trial. Participants were encouraged to regularly view and interact with the posted content throughout the duration of the trial (Figure 1 shows posts). Recommended strategies for effective wall posts (ie, posts that encourage group-member engagement via likes and comments) were utilized throughout the 8 week program [39].

Table 1. Example of posted content during a typical week.

Day and time	Category	Title/"content"	Source
Monday am	Quotation	"Life is like riding a bicycle. In order to keep your balance, you must keep moving."	Albert Einstein
Monday pm	Website	WebMD FIT Teens: Move	WebMD
Tuesday am	Infographic	The Role of Schools in Promoting Physical Activity	Active Living Research
Tuesday pm	Local	Champaign-Urbana Area Bike Routes	Champaign County Bikes
Wednesday am	Mobile app	Walking Paths App	American Heart Association
Wednesday pm	Video PSA	Sedentary-2012	American Academy of Orthopaedic Surgeons
Thursday am	Miscellaneous	We Need More Physical Education in Schools	SPARK
Thursday pm	Website	TeensHealth: Nutrition & Fitness Center	Nemours
Friday am	Local	Hiking in East-Central Illinois	Illinois Department of Natural Resources
Friday pm	Video PSA	ParticipACTION 2012: Driveway	ParticipACTION
Saturday am	Quotation	"Success isn't how far you got, but the distance you traveled from where you started."	Steve Prefontaine
Saturday pm	Infographic	Children and Nature: Being Active in Nature Makes Kids Healthier	National Environmental Education Foundation
Sunday am	Mobile app	Instant Heart Rate App	Azumio
Sunday pm	Miscellaneous	Piano Stairs	The Fun Theory

Figure 1. Screenshot of SMART Facebook Group post. PSA=public service announcements.

Behavioral Condition

Participants in the Behavioral condition received full access to content posted on the *SMART* Group wall and were encouraged to regularly view and interact with the posts. In addition to group access, participants in the Behavioral condition regularly received study-specific behavioral modules via Facebook Messages in the form of 5-10 minute YouTube videos. These video-based modules focused on key elements of physical activity and theoretically based strategies for increased participation among youth. Over the course of the intervention, the group administrator privately delivered eight unique modules to each participant and corresponding guardian on a weekly basis (see Table 2). Along with a link to each weekly module, these messages included a personalized greeting and written information regarding the content of the module.

Table 2. List and location of weekly behavioral modules.

Week number	Module topic
1	Getting Started with the SMART Program
2	Physical Activity Definitions and Benefits
3	Physical Activity Guidelines
4	Goal-Setting for Physical Activity
5	Individual Expectations and Physical Activity
6	Social Support for Physical Activity
7	Overcoming Barriers to Physical Activity
8	Maintaining a Physically Active Lifestyle

Informational Condition

Participants in the Informational condition also received full access to the *SMART* Group and were similarly encouraged to regularly view and interact with the content posted on the group wall. The group administrator via a private Facebook Message also contacted participants once a week. The frequency of these fairly generic messages (ie, weekly greetings) occurred once per week, in unison with the delivery of the behavioral modules in the experimental condition.

Baseline Visit

During their baseline visit to the laboratory, participants, and at least one of their legal guardians, were added to the private *SMART* Group by the group's administrator. Participants and their guardians had to first accept a "Friend Request" from the administrator in order to be electronically invited to join the *SMART* Group. To ensure confidentiality and improve experimental rigor, this group was kept private and was restricted to the randomized child-parent pairs. Once accepted to the *SMART* Group, all participants, regardless of treatment

allocation, were asked (via Facebook Messages) to complete a Web-based battery of questionnaires, prior to the official start date of the trial.

The baseline laboratory visit also included assessments of participants' height and weight. Once these data were obtained, participants were provided with an accelerometer to wear over the course of the following week. A corresponding log to validate wear time and a self-addressed, prestamped envelope to mail back the device were also provided during this visit. Finally, prior to leaving the baseline appointment, all participants, regardless of group allocation, were given pedometers to use over the course of the trial to aid in self-monitoring and behavioral regulation. Following the completion of all baseline data, participants were matched by age and sex and randomized into either the Behavioral or Informational treatment conditions.

Measures

Assessments were conducted at baseline and at the end of the trial (ie, Week 8). All self-report assessments were administered electronically via a Web-based survey service (ie, SurveyMonkey; Palo Alto, CA, 2012). When appropriate, participants received a Web link to a battery of questionnaires via Facebook's direct messaging service. Submitted data were unidentifiable and stored in a secure, password protected online database.

Demographics

Demographic information obtained for the adolescent participants included sex, age, grade level, race, ethnicity, number of siblings, annual household income, and involvement in free or reduced price lunch programs. Parental guardians provided this information during the screening process, and also reported their living situation, highest level of education, and current employment status.

Anthropomorphic Measures

Height and weight were assessed in the laboratory and measured to the nearest 0.1 inches and 0.1 pound, respectively, by using a digital column scale with stadiometer (model 736; Seca, Hamburg, Germany). From these values, participants' body mass index (BMI) was calculated and then interpreted using age-specific BMI percentiles to classify weight status [40].

Objective Physical Activity

Accelerometry was used to objectively assess participants' physical activity levels. Specifically, the rechargeable, lithium-powered Actigraph accelerometer (models GT1M and GT3X; Health One Technology, Fort Walton Beach, FL) was used for this purpose, as it is the most commonly used accelerometer in the field of physical activity-related research [41], and has been shown to provide reliable and valid estimates of energy expenditure and activity levels in youth [42-44]. Participants were instructed to wear the accelerometer on their nondominant hip during all waking hours (with the exception of water-based activities) for seven consecutive days, as recommended by Trost et al [45]. Recommended cut points for predicting physical activity in youth were utilized to properly identify the amount of engagement in sedentary, light, moderate, and vigorous activities [41].

Subjective Physical Activity

Self-reported involvement in physical activities was collected and assessed using the Godin Leisure Time Exercise Questionnaire (GLTEQ) [46]. The GLTEQ is a well validated brief assessment of usual leisure-time exercise habits [47]. Participants were asked to report how many times they participate in strenuous (ie, heart beats rapidly), moderate (ie, not exhausting), and mild (ie, minimal effort) activities for more than 15 minutes over the course of a typical week (ie, seven day period). The reported frequencies of strenuous, moderate, and mild activities are multiplied by 9, 5, and 3 metabolic equivalents, respectively, and then summed to provide a reliable estimate of total and moderate-to-vigorous weekly leisure-time physical activity.

Sedentary Behaviors

The Adolescent Sedentary Activity Questionnaire [48] was used to assess average weekly sedentary behaviors outside of school. Questions about activities normally done while sitting or lying down during a typical week and weekend were asked, and included activities such as "Watching TV", "Using the computer for doing homework", and "Sitting around (chatting with friends/on the phone/chilling)". Participants were asked to report their average time spent engaging in each of these activities for each day of the week.

SMART Facebook Group Usage

Creators and administrators of Facebook groups are provided with basic data regarding how many (and which) group members viewed each post. Following the completion of the trial, the frequency in which each participant viewed the posted content on the *SMART* Group wall was summed and then divided by 120 (ie, total wall posts over the 8 week program) to determine the percentage of content viewed. Additionally, the rate of engagement (ie, the frequency of likes, comments, and shares divided by 120) was immediately calculated following the 8 week program.

Program Evaluation and Feedback

Following the completion of the trial, participants completed an evaluation form regarding the strengths and weaknesses of the *SMART* Trial. There were eight questions regarding participant experience in the *SMART* Trial that were evaluated on 5-point Likert scales. Questions included: (1) "How would you rate your overall experience with the *SMART* Program?"; (2) "How interesting was the content posted on the *SMART* Group wall?"; (3) "How useful was the content posted on the *SMART* Group wall?"; (4) "On average, how many times per week did you visit the *SMART* Group?"; (5) "On average, how often did you interact with the posted content?"; (6) "How much did you learn about physical activity from this program?"; (7) "To what degree did your participation influence your physical activity?"; and (8) "How would you rate your interactions with program staff on Facebook and in person?". Participants were also asked to rate each category of wall posts in order starting with their most favorite (ie, 1) to their least favorite (ie, 7).

Statistical Analysis

Data were checked for missing items, normality, outliers, and errors. A series of independent-samples *t* tests were conducted between groups to identify significant differences in demographic data and descriptive statistics of study variables at baseline. Using an intent-to-treat approach, the potential of the intervention in producing behavioral changes was examined using a 2 (condition, Behavioral vs Informational group) by 2 (time) repeated measures design from data collected at baseline and at the end of the intervention. Interactions and main effects were examined, as well as mean differences in effect sizes. Due to the pilot nature of this trial and the importance of adequately powering subsequent trials, effect sizes (ie, Cohen's *d*) were calculated to further examine the potential value of using social media to successfully deliver a physical activity intervention.

Ethics and Informed Consent

A university Institutional Review Board (Urbana, IL, USA; Protocol No. 13019) approved the study protocol. Upon meeting eligibility criteria, participants received a letter inviting them to participate, a detailed map and information regarding their baseline appointment, and informed consent and assent documents (along with copies of both forms for personal records). At least one parental guardian was required to provide written consent to allow their child to participate in the trial, and the adolescent participant(s) were asked to read and sign an informed assent document prior to being randomized. These documents were similar in language and content and included: (1) a concise overview of the trial and its purpose; (2) an explanation of group randomization and condition-specific expectations; (3) information regarding scheduled onsite appointments, including a description of assessments to be performed at both baseline and follow-up; (4) statements regarding the potential risks and benefits associated with participation in a physical activity trial; (5) a section highlighting participants' rights and privacy, which ensures confidentiality and stresses the voluntary nature of study involvement; (6) confirmation that participation in the study is free; and (7) contact information for the principal investigator and study staff, as well as the university's Institutional Review Board.

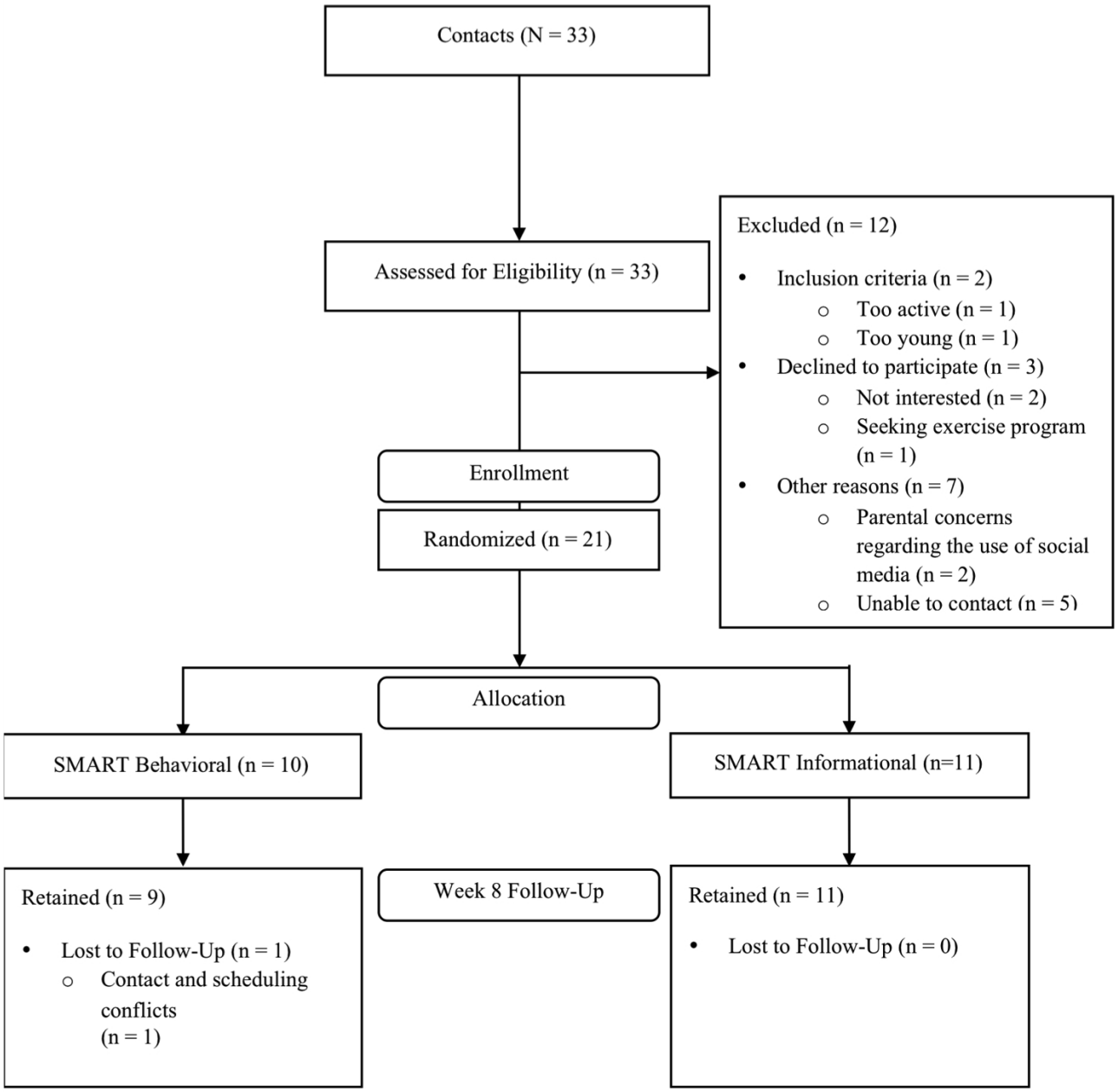
Due to the sensitive nature of conducting research on adolescents, particularly in a Web-based setting, several additional steps were taken to safeguard the rights and welfare of this group. First, all identifiable information was kept in a secure, password protected database and locked filing cabinet separate from participant data, which were coded and aggregated. Second, during the screening process, the legal guardians of all potential participants were required to complete a health history questionnaire, as well as a preparticipation health screening form to ensure safe participation of each child. These procedures allowed us to screen out individuals whose physical condition contraindicates involvement in a physical activity program (n=0). Next, once eligibility was determined and consent was provided, both the adolescent participants and their legal guardians were invited to join the restricted access, study-specific Facebook group via the group's administrator (ie, study staff). Prior to the official start of the program, the only content that was posted on the group wall were links to Facebook's Family Safety Center and Google's Safety Center for Families. These sites provided parent-child dyads with the necessary information and tips needed for using both Facebook and the Internet safely and appropriately. Finally, information shared on the personal Facebook profiles of participants and guardians was not collected nor distributed by the research staff for any purpose whatsoever.

Results

Recruitment and Study Flow

In total, 33 contacts were made and screened for eligibility, but only 21 participants met the inclusion criteria. To ensure equal allocation of subgroups to each treatment condition, eligible participants were randomized after blocking on sex and age (ie, potential confounders) via SPSS version 22 (SPSS IBM). Of the 21 randomized participants, 20 completed assessments at both baseline and follow-up, resulting in 4.8% rate of attrition (Figure 2 shows this).

Figure 2. SMART Trial CONSORT diagram.



Sample Characteristics

Participant characteristics for the full study sample as well as each condition are presented in Table 3. Briefly, a majority of the sample was white (13/21, 61%), female (11/21, 52%), 13 years of age (13/21, 61%; estimated marginal mean age 13.48), in middle school (16/21, 76%), clinically overweight or obese (12/21, 57%), resided with siblings at home (20/21, 95%), and lived in a household with annual income of greater than US $100,000 (15/21, 71%). Additionally, parental characteristics revealed that (21/21) 100% of the sample resided with their mothers, a majority of whom had a bachelor's degree or higher (17/21, 81%) and were employed full time (14/21, 66%). A series of independent-samples *t* tests revealed no significant differences in demographic variables between groups for either the participants or their legal guardians.

Table 3. Participant characteristics.

Variables	Total sample N=21, n (%)	Behavioral group n=10, n (%)	Informational group n=11, n (%)
Sex			
Male	10 (48)	6 (60)	4 (36)
Female	11 (52)	4 (40)	7 (64)
Age (years)			
13	13 (62)	6 (60)	7 (64)
14	6 (29)	3 (30)	3 (27)
15	2 (9)	1 (10)	1 (9)
Year in school (grade)			
6th	1 (5)	0 (0)	1 (9)
7th	6 (29)	3 (30)	3 (27)
8th	9 (43)	5 (50)	4 (37)
9th	3 (14)	1 (10)	2 (18)
10th	2 (9)	1 (10)	1 (9)
BMI classification			
Underweight	1 (5)	0 (0)	1 (9)
Healthy weight	8 (38)	5 (50)	3 (27)
Overweight	3 (14)	1 (10)	2 (18)
Obese	9 (43)	4 (40)	5 (46)
Siblings	20 (95)	10 (100)	10 (90)
Race			
White	13 (62)	6 (60)	7 (64)
Black	1 (5)	1 (10)	0 (0)
Asian	2 (9)	0 (0)	2 (18)
Biracial	5 (24)	3 (30)	2 (18)
Latino	2 (9)	1 (10)	1 (9)
Annual household income			
US $10-40 K[a]	4 (19)	3 (30)	1 (9)
US $41-70 K[a]	1 (5)	0 (0)	1 (9)
US $71-100 K[a]	1 (5)	1 (10)	0 (0)
US >$100 K[a]	15 (71)	6 (60)	9 (82)
Free/reduced price lunch	2 (9)	1 (10)	1 (9)

[a] K=thousand

Intervention Effects on Physical Activity

Independent-samples *t* tests were conducted for the descriptive variables to identify significant differences between the Behavioral and Informational conditions at baseline. At baseline, the only variable that was found to be significantly different between groups was the average amount of time spent engaging in vigorous leisure-time physical activity, where the Informational condition reported a higher rate of engagement (mean 42.55, SD 21.45) in comparison to the Behavioral condition (mean 37.60, SD 15.11); t_{17}= -2.35, *P*=.03.

Next, a series of mixed model analysis of variance (ANOVAs) were performed using a 2 (treatment, Behavioral and Informational conditions) by 2 (time, baseline and Week 8) repeated measures design to examine the effectiveness of the intervention in producing changes in physical activity behaviors (see Table 4). Analyses revealed that there were significant improvements over time on subjectively reported weekly leisure-time physical activity, but there was no interaction

between time and condition. Changes in subjectively reported moderate-to-vigorous physical activity approached significance over time, but, again, significant interaction effects were not found. Furthermore, there were no significant time effects among the objectively measured physical activity variables, including average daily minutes spent being physically active, sedentary time, moderate-to-vigorous physical activity, and total physical activity. Similarly, significant group by time effects were not present for any of the objectively assessed variables. Finally, analyses of self-reported time spent engaging in weekday and weekend sedentary behaviors did not produce significant time or interaction effects.

Given the small sample size of this trial, effect sizes (ie, Cohen's *d*) were calculated to identify the patterns of change for the total sample and within each treatment group. Mean values for physical activity and sedentary outcomes as well as effect sizes are reported in Table 5. Changes in the behavioral outcomes were generally moderate in size and in the expected direction.

Table 4. Time and interaction effects for behavioral outcomes.

	Time effects				Time X group effects			
Variables	M [a]	$F_{1,18}$	P	η^2	M [a]	$F_{1,18}$	P	η^2
Subjective physical activity								
MVPA[b] leisure-time	47.250	4.186	.056	0.189	46.949	0.029	.868	0.002
Total leisure-time	57.900	8.426	.009	0.319	57.465	0.002	.968	0.000
Objective physical activity								
MVPA[b] counts	179.539	0.649	.435	0.048	179.199	1.045	.325	0.074
Total counts	279,923.933	1.493	.243	0.103	279,238.415	0.631	.441	0.046
Sedentary behavior								
Weekday	56.464	0.445	.513	0.024	56.744	0.118	.735	0.007
Weekend	65.838	0.068	.798	0.004	65.881	0.057	.814	0.003

[a]Estimated marginal means

[b]MVPA = moderate-to-vigorous physical activity

Table 5. Descriptive statistics and effect sizes of study variables.

Variables	Baseline mean (SD)	Follow-up mean (SD)	Effect size Cohen's *d*
MVPA [a] leisure-time			
Behavioral	37.60 (15.11)	50.22 (14.08)	0.86
Informational	42.55 (21.45)	57.36 (37.79)	0.50
Total sample	40.19 (18.42)	54.15 (29.12)	0.59
Total leisure-time			
Behavioral	43.00 (17.66)	62.56 (17.21)	1.12
Informational	52.64 (21.50)	71.00 (34.65)	0.65
Total sample	48.05 (19.90)	67.20 (27.84)	0.80
MVPA [a] counts			
Behavioral	152.55 (33.30)	189.74 (53.73)	0.85
Informational	177.69 (83.73)	182.45 (66.51)	0.06
Total sample	165.71 (64.57)	185.85 (58.85)	0.33
Total counts			
Behavioral	235,884.95 (56,843.58)	293,563.02 (86,377.37)	0.81
Informational	276,452.27 (119,017.51)	294,741.53 (108,012.65)	0.16
Total sample	257,134.50 (94,697.62)	294,191.56 (95,033.31)	0.39
Sedentary weekday			
Behavioral	61.18 (21.39)	57.68 (26.02)	-0.15
Informational	54.54 (19.22)	53.34 (20.54)	-0.06
Total sample	57.70 (20.05)	55.30 (22.63)	-0.11
Sedentary weekend			
Behavioral	67.65 (38.01)	65.25 (34.85)	-0.07
Informational	65.49 (19.68)	65.40 (22.19)	0.00
Total sample	65.12 (29.07)	65.33 (27.76)	0.01

[a]MVPA = moderate-to-vigorous physical activity

Program Engagement and Feedback

Following program completion, participant use and engagement were assessed. Briefly, (96/120) 80.0% of the daily posts made on the *SMART* Group wall were viewed by the total sample. This rate was notably higher in the Behavioral condition (104/120, 86.6%) than in the Informational condition (88/120, 73.3%), however this difference was not statistically significant. Group engagement (ie, likes, comments, and shares), on the other hand, was relatively low among the total sample (32/120, 26.7%), and similar between the Behavioral and Informational conditions (ie, 33/120, 27.5% and 31/120, 25.8%, respectively).

Participants were asked to anonymously complete a program evaluation and feedback form that was designed specifically for this trial. There were 20 of the 21 participants completed these forms at their follow-up appointment. Results revealed that (14/20) 70% of the study sample was "satisfied" to "very satisfied" with their overall experience with the *SMART* Trial. Additionally, (11/20) 55% of the sample found the content posted on the *SMART* Group wall to be "interesting" to "very interesting", and (9/20) 45% found the posted content to be "useful" to "very useful". On average, (11/20) 55% of the participants visited the *SMART* Group "1-2 times per week", while (9/20) 45% interacted with the posted content "1-2 times per week", as well. Furthermore, (10/20) 50% of those surveyed indicated that they learned "a good amount" to "a great deal/a lot" about physical activity from their involvement in this program, and (14/20) 70% felt that their participation in the program influenced their physical activity "some" to "a good amount". And finally, when asked to rate their experiences with study staff (over Facebook and in person), (19/20) 95% of the participants reported having "good" to "excellent" interactions. Insight into participants' preferred categories of wall posts was also assessed, with the Technology/App category being the most preferred type of shared content, while Infographics were rated as the least preferred category (see Table 6).

Table 6. Average ratings of wall post categories.

Category	Mean[a] (SD)
Infographics	4.84 (2.39)
Local	4.26 (1.63)
Quotation	3.89 (1.82)
Technology	2.89 (1.88)
Video PSA	4.58 (2.09)
Websites	3.05 (1.78)
Miscellaneous	3.79 (1.87)

[a] Lower means indicate more favorable ratings

Discussion

SMART Trial Objectives

The purpose of the *SMART* Trial was to examine the feasibility of using social media to deliver a physical activity intervention to low-active adolescents 13 to 15 years old. An additional aim of this trial was to examine the degree of changes in behavioral outcomes within the whole sample, as well as both treatment conditions.

Primary Outcomes

According to results from the ANOVAs, participation in total leisure-time physical activity was the only behavioral outcome to positively and significantly change over time. Furthermore, significant group by time interactions were not present for any of the behavioral outcomes assessed. Despite the lack of statistically significant findings, however, involvement in this trial did result in small increases in objectively assessed moderate-to-vigorous and total physical activity, as well as time spent being physically active. Similarly, subjectively assessed physical activity resulted in improvements, but the effects were larger for moderate-to-vigorous physical activity (MVPA) and total weekly leisure-time physical activity. Although both forms of measurement are intended to capture one's level of physical activity, it is important to note that the GLTEQ was specifically designed to assess *leisure-time* physical activity behaviors [46], whereas the accelerometer assesses *all* physical activity accumulated throughout the day, regardless of activity type. Therefore, participation in this trial appeared to be more beneficial for influencing leisure-time physical activities, such as going to the park or walking the dog, than for total daily physical activities, such as getting ready for school or doing chores.

Due to the lack of statistical power to reveal conventional differences between treatment conditions, effect sizes were calculated to further examine changes in the behavioral outcomes. Effect sizes for both the subjectively (ie, MVPA and total leisure-time physical activity) and objectively (ie, MVPA and total counts) assessed physical activity variables were, in most cases, moderate to large in size and in the hypothesized direction for the study sample. Conversely, changes in self-reported sedentary behaviors were small to nonexistent. This is not entirely surprising, however, as the purpose of this trial was not aimed at minimizing sedentary behaviors, but, rather, increasing regular participation in physical activities.

It should be noted that the effect sizes for physical activity in this particular trial were generally larger than those reported in the literature. For example, Kamath et al conducted a meta-analysis on behavioral interventions aimed at improving physical activity levels in children and adolescents, noting that youth-based physical activity interventions typically result in a significant, but small effect (d=0.12; range, 0.04-0.20) [49]. Moreover, Lau et al conducted a review of information and communication technology-based interventions and also found small effect sizes, ranging from 0.03 to 0.41, always favoring the intervention over the control conditions [24]. However, none of the interventions in these reviews were entirely delivered via social media. The social and interactive nature of the *SMART* Trial, in conjunction with the weekly receipt and viewing of the video-based behavioral modules, may provide some insight as to why this social media-delivered intervention resulted in larger effects than similar programs, which have preceded it.

Although these results appear to be promising, they should be interpreted with caution. The effect size results are helpful in determining the degree of influence that program involvement may have had on physical activity behaviors, but there remained an inability to detect significant differences between the treatment conditions via inferential statistics. Moreover, the limited power and lack of a true control group (ie, not receiving any form of intervention throughout the study period) limits the ability to adequately assess whether or not the addition of the behavioral modules, in particular, increased the efficacy of this social media-delivered intervention and contributed to changes in physical activity. The changes that occurred in the total study sample or by condition could have also been the result of many other factors including, but not limited to, the provision of pedometers, parental support, and allotted screen time. At the very least, results from these secondary analyses provide additional support for the need to further investigate this novel approach of delivering behavioral interventions via social media.

Finally, the design and delivery of the *SMART* Trial resulted in a relatively high rate of program engagement among study participants in both treatment conditions. This may have been the result of continually changing, yet focused content shared in a virtual community of similar others, as well as following guidelines for effective wall posts [39]. Furthermore, participants

were given blatant instructions and reminders to regularly view and interact with the posted content on a daily basis. It should be noted, however, that passive engagement (ie, viewing shared content) was greater than active engagement (ie, likes, comments, and shares). This unique finding warrants further investigation, as passive engagement may simply serve as an indicator of program adherence, whereas active engagement may be indicative of participants' interests in and preferences for select content. Whether or not these types of virtual interactions can translate to meaningful, real-world changes in health behaviors, such as physical activity, remains to be determined.

Strengths

To our knowledge, this is the first pilot trial examining the feasibility of delivering a randomized controlled physical activity intervention entirely via social media, and specifically targeting the understudied demographic of early adolescence. The use and comparison of objective and subjective assessments of physical activity also reflect strengths of the study, as multiple forms of measurement can provide greater accuracy of and insight to physical activity behaviors among youth [50]. Finally, participants reported a high degree of satisfaction with their participation in this program, indicating that the posted content was interesting, informative, and useful. Therefore, delivering a behavioral intervention over social media appears to be a practical and well accepted approach to encouraging youth to become more physically active.

Limitations

Several limitations with this trial should be considered when interpreting the results and building upon the findings. First, despite considerable efforts in trying to recruit participants for this trial, the study sample was relatively small. A larger sample size would improve the power of the study and potentially reduce the amount of variance among reported outcomes. Additionally, a majority of the sample came from higher socioeconomically status households. Whether or not similar results would be found in participants from lower socioeconomic households remains to be determined. A further limitation was the lack of ability to track the viewership of the weekly behavioral modules by participants (ie, the only difference between the two treatment conditions). Examination of the compliance and frequency of viewership could have provided a more concrete conclusion regarding the overall utility and worth of these modules in promoting positive changes in physical activity behaviors. Finally, issues regarding recall and reporting biases should be taken into consideration when interpreting results obtained from subjective assessments.

Future Directions

Research utilizing and examining the effectiveness of social media in improving health behaviors and outcomes is still in its nascent stage and requires further investigation [51]. While the larger than average effect sizes found in this pilot trial are encouraging, larger studies should be conducted to better determine the effectiveness of using social media to promote physical activity behaviors in low-active adolescents. If efficacy can be established, similar trials should be conducted and evaluated with other populations of varying ages and perhaps even disease states. It is also important to examine participants' abilities to maintain improved levels of physical activity over time, particularly as shared information and behavioral strategies may remain readily accessible following the end of such an intervention via continual access to and further engagement with previously posted content following program termination. Relative to the evaluation of social media as a form of treatment delivery, researchers should further investigate the differential influence, if any, of passive and active engagement in a virtual environment on physical activity behaviors. Additionally, new and innovative ways to increase engagement among participants should be identified and evaluated. For example, encouraging more participant/user-generated content, such as sharing photos of places to be active or creating "how to" video tutorials, may increase perceptions of ownership and accountability among participants in the program. Indeed, increased engagement by participants should lead to more effective interventions delivered via social media, and, as a result, may lead to greater program satisfaction as well as improved health-related behaviors and outcomes [51]. Last, while the *SMART* Trial chose to use Facebook to deliver this intervention, future studies may want to explore the potential of other commonly used social media services (eg, Twitter) to examine and compare their potential to promote health-related behaviors [52].

Conclusions

The present study provides initial support for the feasibility of delivering behavioral interventions via social media [53]. The social and interactive nature of social media, along with its low cost and accessibility, make it an appealing avenue in which to target and influence health behaviors, such as physical activity. Furthermore, delivering social media-based programs can overcome many of the constraints that are commonly found with more traditional Internet- (eg, limited interactivity), print- (eg, text-heavy), and/or center-based (eg, travel) interventions. However, the effectiveness of promoting positive behavior change via social media remains to be determined (see Multimedia Appendix 1).

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Conflicts of Interest

None declared.

Multimedia Appendix 1

PowerPoint slides for conference presentation.

[PDF File (Adobe PDF File), 3MB - resprot_v3i4e56_app1.pdf]

Multimedia Appendix 2

CONSORT-EHEALTH checklist V1.6.2 [54].

[PDF File (Adobe PDF File), 995KB - resprot_v3i4e56_app2.pdf]

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Abbreviations

ANOVA: analysis of variance
BMI: body mass index
GLTEQ: Godin Leisure-Time Exercise Questionnaire
MVPA: moderate-to-vigorous physical activity
PSA: public service announcement
SMART: Social Media and Activity Research in Teens

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Original Paper

A Web-Based Training Program Using Cognitive Behavioral Therapy to Alleviate Psychological Distress Among Employees: Randomized Controlled Pilot Trial

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Abstract

Background: A number of psychoeducational programs based on cognitive behavioral therapy (CBT) to alleviate psychological distress have been developed for implementation in clinical settings. However, while these programs are considered critical components of stress management education in a workplace setting, they are required to be brief and simple to implement, which can hinder development.

Objective: The intent of the study was to examine the effects of a brief training program based on CBT in alleviating psychological distress among employees and facilitating self-evaluation of stress management skills, including improving the ability to recognize dysfunctional thinking patterns, transform dysfunctional thoughts to functional ones, cope with stress, and solve problems.

Methods: Of the 187 employees at an information technology company in Tokyo, Japan, 168 consented to participate in our non-blinded randomized controlled study. The training group received CBT group education by a qualified CBT expert and 1 month of follow-up Web-based CBT homework. The effects of this educational program on the psychological distress and stress management skills of employees were examined immediately after completion of training and then again after 6 months.

Results: Although the training group did exhibit lower mean scores on the Kessler-6 (K6) scale for psychological distress after 6 months, the difference from the control group was not significant. However, the ability of training group participants to recognize dysfunctional thinking was significantly improved both immediately after training completion and after 6 months. While the ability of participants to cope with stress was not significantly improved immediately after training, improvement was noted after 6 months in the training group. No notable improvements were observed in the ability of participants to transform thoughts from dysfunctional to functional or in problem-solving skills. A sub-analysis of participants who initially exhibited clinically significant psychological distress (K6 score ≥5) showed that the mean K6 score was significantly improved immediately after training completion for the training group compared to the control group (−2.50 vs −0.07; mean difference 2.43, 95% CI 0.55-4.31; d=0.61), with this effect remaining even after 6 months (−3.49 vs −0.50; mean difference 2.99, 95% CI 0.70-5.29; d=0.60).

Conclusions: Our results suggest that a brief stress management program that combines group CBT education with Web-based CBT homework moderately alleviates the distress of employees with clinically significant psychological distress. In addition, the program might help improve employees' ability to evaluate their own stress management skills.

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KEYWORDS

Web-based training program; cognitive behavioral therapy; stress management; workplace; Internet; group

Introduction

Alleviating psychological distress in employees is essential in terms of health and work performance. Epidemiological studies have reported that the proportion of workers whose stress level is high enough to necessitate treatment is approximately 15% [1,2]. Despite this, many experiencing such high levels of stress continue to work without receiving proper care [1]. According to a Japanese national survey, 58% of workers report feeling significantly strong psychological distress [3].

Systematic reviews have reported that work-related psychological distress, regardless of clinical significance, has been found to be related to a number of mental disorders, including depression and anxiety disorders [4,5]. Further, to improve work performance, increasing emphasis is being placed on managing psychological distress regardless of clinical significance [6,7].

In a national survey, 60% of workers reported strong psychological distress, which suggests that other workers might also feel some degree of psychological distress, the Japanese Ministry of Health, Labor and Welfare has encouraged employers to implement stress management education for all employees to prevent mental disorders and improve work performance [8].

A systematic review of the effects of training programs for work-related stress reported that those using cognitive behavioral therapy (CBT) is the most effective option for alleviating stress [9]. These training programs require time and CBT experts and are conducted not only as therapy but also as stress management training [10-20]. CBT has mainly been implemented as an individual psychotherapy, although low-intensity CBT [21-23] to provide interventions to a relatively larger number of people is becoming increasingly common. Recent programs include the provision of information on CBT via books, group training, and group education. CBT is now also offered via telephone and Web in a self-learning format. This increase in accessibility has proven beneficial in improving stress management [9,24-28]. However, despite these promising findings regarding CBT, its efficacy in the workplace remains uncertain, as the time available for health education of employees in a workplace setting is limited.

Here, we developed a brief educational program based on CBT that is feasible for implementation in a workplace setting. We conducted a pilot study with a randomized controlled design to investigate the effects of the program on alleviation of distress and improvement of stress management abilities in employees who have significant or non-significant distress.

Methods

Study Participants and Procedure

The target population consisted of 187 employees (147 men and 40 women) at an information technology company in Tokyo, Japan. The majority of employees were system engineers with a high degree of computer literacy. The company provides in-house training programs for managerial (eg, legal knowledge, human resource management, accounting) and non-managerial positions (eg, health and safety) once or twice a year. The present training program was announced and briefed for the 187 non-management employees in the company. Participation in this study was voluntary, and informed consent was obtained from employees prior to group education after explaining the study purpose, procedures, and details of the training program.

The participants who provided written informed consent were randomly assigned to training or control groups. The training group received group CBT training during working hours in December 2011 and an additional month of CBT homework, with a follow-up study conducted 6 months later. All participants were required to complete self-rated online questionnaires before training, immediately after, and on 6-month follow-up. For ethical reasons, this training program was provided to the control group after follow-up. No exclusion criteria were set, as the study examined the effects of the CBT program in a real-world workplace setting. The study protocol was approved by the Ethics Committee of the School of Allied Health Sciences at Kitasato University. Reporting of methods and results of this study are based on the CONSORT-EHEALTH guidelines [29].

Contents of the Training Program

The training program was composed of a 150-minute group class presented by a qualified CBT expert on cognitive behavior therapy and 1 month of homework via Web-based CBT program. The following three topics were covered in the group education program: an overview of CBT, problem-solving techniques, and cognitive restructuring techniques. For problem-solving techniques, we encouraged participants to use group brainstorming, which we consider the most important element in a number of problem-solving techniques. For cognitive restructuring techniques, participants received a lecture designed to facilitate understanding of the techniques and column sheets to complete during the Web-based CBT program. Column sheets included the following topics: situations where participants felt stressed, feelings and behaviors, automatic thoughts, an objective examination of those automatic thoughts (including counterevidence), adaptive thoughts, and changes in feeling and behavior. While completing the worksheets, participants held group discussions by exchanging questions and opinions regarding the cognitive restructuring techniques. Participants also improved their understanding of the cognitive restructuring techniques by consulting with the CBT expert. Following group education, the Web-based CBT homework was explained.

Participants were asked to practice the column method with a Web-based CBT program by reflecting on the stress they experienced over a 1-month period starting the day after group education. The Web-based CBT program was developed by Woman Wave Corporation (see Figures 1-2 for screenshots). The fee for the program was prepaid using research expenses

to allow participants to access the program free of charge. The recommended Web program enables users to easily complete the column sheets by providing contextual explanations and advice on the column method. Users can complete column sheets by entering information as directed on the screen. In addition to the column sheet above, participants were able to access the program via their computers at work and home and were encouraged to complete their homework at least twice. Homework was expected to help familiarize participants with the column method. However, homework completion was not mandatory, to avoid placing further pressure on participants.

To encourage homework completion, occupational health nurses sent a total of four emails, prepared by a CBT expert, to each participant to provide supplementary information and tips regarding the cognitive restructuring techniques. These occupational health nurses received a 150-minute training session before the study from a CBT expert and then answered questions from participants regarding Web-based CBT procedures, with a CBT expert answering any remaining questions. To reduce the burden on participants, the program did not contain homework on problem-solving techniques.

Figure 1. Program screenshot.

Figure 2. Program screenshot.

Outcome Evaluation

The primary outcome was measured as the change in Kessler-6 (K6) score, which measures psychological distress. The K6 score was measured before training to establish a baseline and then immediately after 1 month of training and again after 6 months. Developed as a screening tool for depressive disorders and anxiety disorders [30], the K6 scale is widely used to assess psychological stress [31,32], with the score obtained from a simple self-rating questionnaire on symptoms of depression and anxiety experienced over the previous month. The reliability and validity of the Japanese version of the K6 questionnaire utilized in this study have been verified [33]. A study on the cutoff point to diagnose clinically significant psychological distress of respondents suggest a cutoff point of 4 or 5 (total score, 24) [34].

Secondary outcomes were evaluated based on respondents' answers to several questions on an original self-rating questionnaire. Questions concerned the recognition of dysfunctional thinking habits ("Do you recognize your dysfunctional thinking habits?"), ability to change dysfunctional thinking patterns to functional ones ("Can you transform your dysfunctional thinking patterns that have been bothering you into functional ones?"), ability to cope with stress ("Are you confident that you can cope with stress by yourself?"), and problem-solving skills ("Do you think you can solve a problem when you face one?"). The respondents answered using a 5-point scale (1=strongly disagree, 5=strongly agree).

Randomization and Masking

An independent researcher who had no direct contact with the participants used computer-generated randomization with a 1:1 ratio and block size of 6. No stratification was performed and evaluators were masked. Owing to the nature of the intervention, participants were informed of their allocation status.

Statistical Analysis

A systematic review of the literature on mental disorder intervention suggests that Cohen's effect size (*d*) for those with sub-threshold depression is 0.42 (95% CI 0.23-0.60) [35]. The sample size necessary to obtain an effect size of 0.42 with probability of Type I error (α) less than .05 and Type II error (β) less than .20 was 90 for each group. A generalized equation was used for estimation, based on an intention-to-treat (ITT) analysis. The rate of missing primary or secondary outcomes was 19.26% across the follow-up period (K6, 18.2%; recognition of dysfunctional thinking habits, 19.6%; changing dysfunctional thinking patterns, 19.3%; coping skills, 19.0%; problem-solving skills, 19.6%). To satisfy the ITT requirement that analyses be conducted for all participants, a multiple imputation (MI) method was used on the assumption that data could be considered missing at random. MI allows for uncertainty caused by missing data by generating several different plausible imputed data sets using a set of external covariates and appropriately combining results obtained from each [36,37]. We utilized a sequential regression approach to generate 20 imputations for each missing value, as recommended by Graham [38].

To determine the effects of the training program, primary and secondary outcomes were measured, and differences in scores before and after implementation for the training and control groups were calculated. The short-term effect was calculated by subtracting the baseline scores from those obtained after completion of 1 month of homework. The long-term effect was calculated by subtracting the baseline scores from those obtained after 6 months. Results are shown as changes in the raw scores for primary and secondary outcomes. In addition, the differences in mean adjusted for baseline score of each outcome were also calculated. For the K6 scale, a sub-analysis was conducted among participants with a K6 score ≥5 at baseline. In addition, the training group was divided into subgroups of those completing Web-based CBT homework at least once and those completing no homework, and changes in the K6 scores of these subgroups were compared.

To analyze baseline characteristics of the study participants, information on sex, age, hours of overtime, mean hours of sleep on weekdays, marital status, drinking habits, exercise habits, and history of psychiatric treatment was collected from each participant at baseline. A *t* test was used for numerical variables and a χ^2 test for categorical variables. Statistical significance was set at $P<.05$. IBM SPSS Statistics 22 and IBM SPSS Missing Values 22 (*SPSS* Inc., *Chicago*, IL, USA) were used for statistical analyses.

Results

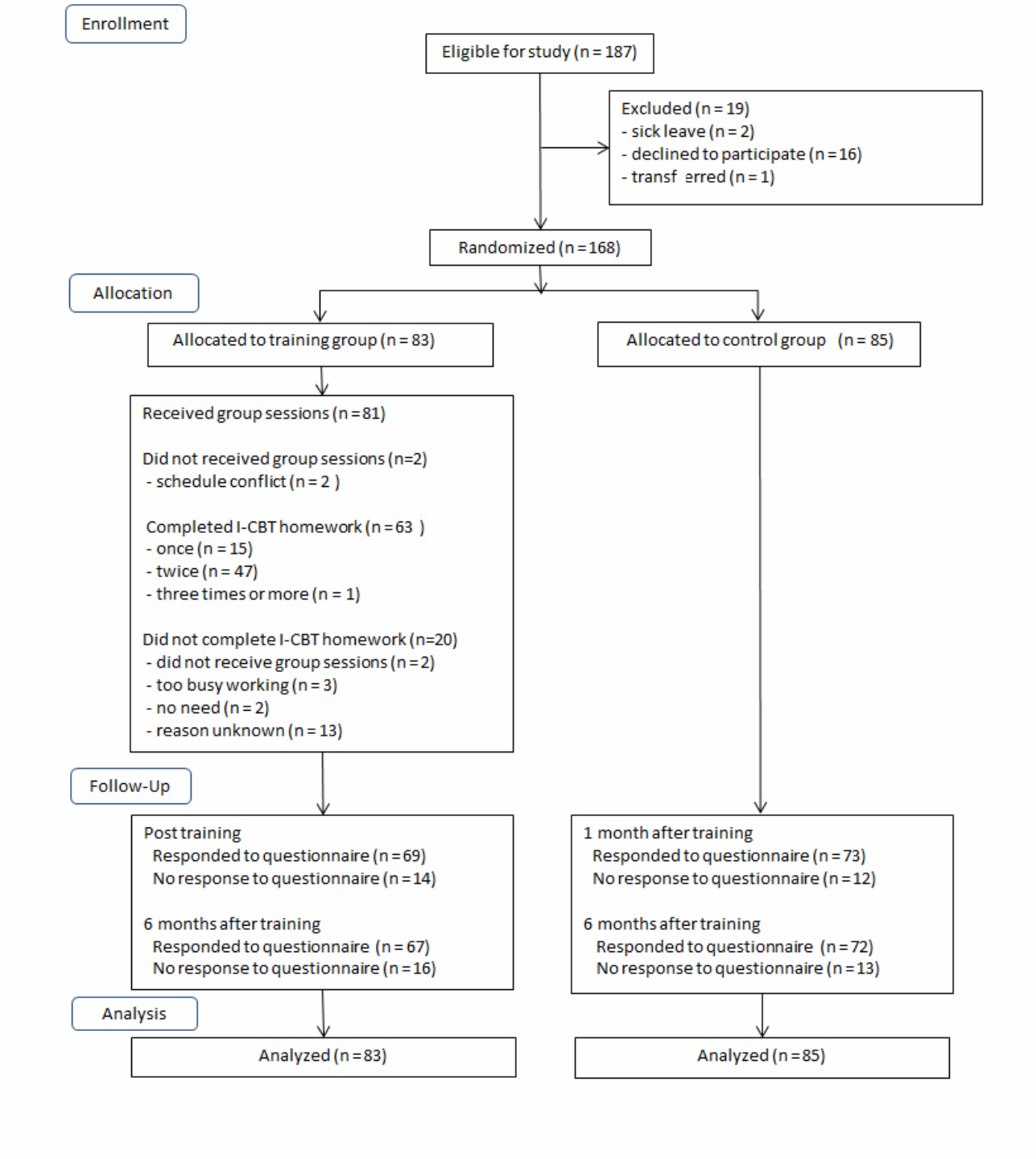
Participants

Figure 3 shows the study flow. Of 187 potential participants, 2 took a leave of absence before the start of the study, 16 did not consent to participate, and 1 was transferred to another location before the study started, leaving 168 enrolled in this study. These 168 participants were randomly assigned to either the training (n=83) or control group (n=85). In the training group, 81 (98%) actually received a group session, and 63 (76%) actually received additional Web-based CBT homework. The completion of homework by participants was as follows: once (n=15), twice (n=47), and more than twice (n=1). Among those who did not complete assigned homework (20/83, 24%), 2 also did not attend the group session.

Follow-up questionnaires immediately after completion of the program were completed by 69 (83%) of the 83 respondents in the training group and by 73 (86%) of the 85 control group participants. Follow-up questionnaires after 6 months were completed by 67 (81%) respondents in the training group and by 72 (85%) in the control group.

Five participants emailed the nurse to inquire about the Web-based CBT. Of these five, two inquired about how to operate the Web program and three about how to complete the column sheets. The content of this advice was the same as that provided in the training session.

Figure 3. Trial profile.



Baseline Characteristics

Baseline characteristics of the 168 participants are shown in Table 1. No missing data were observed. Of the 168 participants, 22% (n=37) were women (19%, 16/83 of training group and 25%, 21/85 of control group), 78% (n=131) were men (81%, 67/83 of training group and 75%, 64/83 of control group), and mean age across both training and control groups was 38.4 years (SD 8.1 and SD 8.4 respectively). No significant differences were observed between the training and control groups regarding sex ratio, mean age, hours of overtime, mean hours of sleep, marital status, exercise and drinking habits, K6 score, ability to recognize and change dysfunctional thinking habits, ability to cope with stress, or problem-solving skills. Although a proportion of participants had pre-existing or ongoing mental health conditions that required psychiatric treatment, this was not significant relative to the populations of the training and control groups. Differences in primary and secondary outcome scores, age, and sex ratio at baseline also did not significantly differ between those who responded to the follow-up questionnaires, both immediately after the study and after 6 months, and those who did not.

Table 1. Baseline characteristics of participants in training and control groups.[a,b]

Characteristic		Total (n=168)	Training group (n=83)	Control group (n=85)	*P* value
Men, n (%)		131 (78.0)	67 (80.7)	64 (75.3)	.26
Age, years, mean (SD)		38.4 (8.3)	38.4 (8.1)	38.4 (8.4)	.35
Overtime hours, n (%)					
	Few	54 (32.1)	28 (33.7)	26 (30.6)	.34
	<40 hours	97 (57.7)	46 (55.4)	51 (60.0)	
	40-79 hours	17 (10.1)	9 (10.8)	8 (9.4)	
	≥ 80 hours	0 (0)	0 (0)	0 (0)	
Mean hours of sleep (weekday), n (%)					
	<5 hours	17 (10.1)	9 (10.8)	8 (9.4)	.84
	5-6 hours	124 (73.8)	59 (71.1)	65 (76.5)	
	7-8 hours	26 (15.5)	15 (18.1)	11 (13.1)	
	≥9 hours	1 (0.60)	0 (0)	1 (1.20)	
Marital status, n (%)					
	Married	77 (45.8)	41 (49.4)	36 (42.4)	.41
	Single	91 (54.2)	42 (50.6)	49 (57.6)	
Drinking, n (%)					
	None	64 (38.1)	33 (39.8)	31 (36.5)	.29
	1-3 days/week	61 (36.3)	26 (19.3)	35 (41.2)	
	4-6 days/week	19 (11.3)	10 (12.0)	9 (10.6)	
	Every day	24 (14.3)	14 (16.9)	10 (11.8)	
Exercise habit, n (%)					
	None	88 (52.4)	41 (49.4)	47 (55.3)	.6
	1-2 times/week	65 (38.7)	35 (42.2)	30 (35.3)	
	≥3 times/week	15 (8.90)	7 (8.40)	8 (9.40)	
History of psychiatric treatment, n (%)					
	No history	143 (85.1)	69 (83.1)	74 (87.1)	.28
	History of treatment	16 (9.50)	8 (9.60)	8 (9.40)	
	Undergoing treatment at present	9 (5.40)	6 (7.20)	3 (3.50)	
K6 score, mean (SD)		4.8 (4.50)	4.7 (4.50)	4.8 (4.50)	.90
Recognition of dysfunctional thinking habits, mean (SD)		2.36 (0.95)	2.53 (1.02)	2.20 (0.87)	.57
Changing dysfunctional thinking patterns, mean (SD)		3.17 (0.89)	3.10 (0.91)	3.24 (0.86)	.33
Coping skills, mean (SD)		3.08 (0.93)	3.10 (0.90)	3.06 (0.96)	.49
Problem-solving skills, mean (SD)		2.87 (0.94)	2.82 (0.97)	2.92 (0.91)	.54

[a]Independent *t* test for difference between groups for continuous measures and chi-square test for differences between groups for categorical characteristics.
[b]Scores on a scale 1-5, with 1 indicating not at all and 5 indicating very well.

Effects of Training Program

Table 2 shows the results of ITT analysis. From before to immediately after training, the mean K6 score of the training group decreased by 0.46 while that of the control group increased by 0.22. However, this difference was not significant (mean difference 0.68, 95% CI −0.44 to 1.80). The intergroup difference in change in mean K6 score for the training and control groups from baseline to 6 months after program completion was also not significant (−0.14 at baseline, 0.83

after 6 months; mean difference 0.97, 95% CI −0.64 to 2.59). No significant effect due to the training program was observed, even when K6 scores were adjusted for baseline scores. Further, while ability of participants to recognize their own dysfunctional thinking was significantly increased both immediately after training (mean difference 0.33, 95% CI 0.06-0.59; *d*=0.37) and at 6 months (mean difference 0.45, 95% CI 0.06-0.83; *d*=0.33), significance was not observed after adjusting for baseline scores of recognition of dysfunctional thinking habits. The ability of participants to transform thought patterns from dysfunctional to functional was also not improved immediately after training in the training group. However, after adjustment for baseline scores, a small significant difference was observed immediately after training, but not after 6 months. While the ability of participants to cope with stress was not significantly improved immediately after training, significant improvement was noted after 6 months in the training group (mean difference 0.54, 95% CI 0.10-0.98; *d*=0.37), a finding that remained even after adjustment for baseline scores. The problem-solving ability of participants was not improved either immediately after training or after 6 months.

At baseline, 36 participants in the training group (43%, 36/83) and 37 in the control group (44%, 37/85) exhibited clinically significant psychological distress (K6 score ≥5). Results of subgroup analysis are shown in Table 3. Mean K6 score was significantly improved in the training group compared with the control group immediately after training (−2.50 vs −0.07; mean difference 2.43, 95% CI 0.55-4.31; *d*=0.61), and this effect remained even after 6 months (−3.49 vs −0.50; mean difference 2.99, 95% CI 0.70-5.29; *d*=0.60) and on adjustment for baseline scores. In addition, the 36 participants of the training group with K6 cutoff score ≥5 points were subdivided into a complete group (n=28) who completed at least 1 homework session and an incomplete group (n=8) who completed no sessions. In contrast to the 37 participants in the control group with a baseline K6 score ≥5, the Web-based CBT complete subgroup had significantly lower mean scores both immediately after training (−2.60 vs −0.07; mean difference 2.53, 95% CI 0.46-4.60; *d*=0.63) and 6 months later (−4.02 vs −0.50; mean difference 3.52, 95% CI 1.13-5.90; *d*=0.74). The same result was observed when scores were adjusted for baseline scores. In addition, mean K6 score for those completing no homework (or Web-based CBT incomplete subgroup) was lower, though not significantly, immediately after training (−2.14 vs −0.07; mean difference 2.07, 95% CI −0.65 to 4.80; *d*=0.53) but increased 6 months later with no difference being observed from the control group (−1.67 vs −0.50; mean difference 1.16, 95% CI −3.44 to 5.77; *d*=0.23). The same result was observed when scores were adjusted for baseline scores.

Study Safety

This study did not exacerbate any existing psychological problems of any participants. While one subject did experience mild distress during group education after being reminded of painful memories, they recovered quickly.

Table 2. Intention-to-treat analyses of primary and secondary outcomes after training and 6-month follow up.

Variable	Immediately after training				6-month follow-up			
	Mean (SE) change	Unadjusted difference in mean (95% CI)	Adjusted difference in mean (95% CI)[a]	Effect size[b]	Mean (SD) change	Unadjusted difference in mean (95% CI)	Adjusted difference in mean (95% CI)[a)]	Effect size[b]
K6 scores								
Training group	−0.46 (0.44)	0.68 (−0.44 to 1.80)	0.70 (−0.34 to 1.73)	0.19	−0.14 (0.64)	0.97 (−0.64 to 2.59)	0.99 (−0.45 to 2.44)	0.18
Control group	0.22 (0.34)				0.83 (0.55)			
Recognizing dysfunctional thinking habits								
Training group	0.40 (0.10)	0.33 (0.06-0.59)[c]	0.19 (−0.02 to 0.40)	0.37	0.68 (0.15)	0.45 (0.06-0.83)[c]	0.26 (−0.04 to 0.56)	0.33
Control group	0.08 (0.09)				0.24 (0.14)			
Changing dysfunctional thinking patterns								
Training group	0.27 (0.08)	0.18 (−0.04 to 0.40)	0.23 (0.02-0.44)[c]	0.26	0.43 (0.20)	0.23 (−0.28 to 0.74)	0.31 (−0.12 to 0.75)	0.13
Control group	0.08 (0.08)				0.21 (0.17)			
Coping skills								
Training group	0.21 (0.10)	0.23 (−0.02 to 0.48)	0.21 (−0.02 to 0.45)	0.27	0.77 (0.16)	0.54 (0.10-0.98)[c]	0.53 (0.15-0.91)[d]	0.37
Control group	−0.01 (0.08)				0.23 (0.16)			
Problem-solving skills								
Training group	0.12 (0.12)	0.17 (−0.13 to 0.47)	0.17 (−0.12 to 0.45)	0.18	−0.01 (0.20)	0.11 (−0.43 to 0.64)	0.06 (−0.41 to 0.54)	0.06
Control group	−0.06 (0.10)				−0.12 (0.19)			

[a]Adjusted for baseline scores.

[b]Unadjusted Cohen's *d*.

[c]*P*<.05.

[d]*P*<.01.

Table 3. Subanalyses of K6 score among participants with K6 ≥5 at baseline.

Subgroup	Immediately after training				6-month follow-up			
	Mean (SE) change	Unadjusted difference in mean (95% CI)	Adjusted difference in mean (95% CI)[a]	Effect size[b]	Mean (SD) change	Unadjusted difference in mean (95% CI)	Adjusted difference in mean (95% CI)[a]	Effect size[b]
Training group (all), n=36	−2.50 (0.67)	2.43 (0.55-4.31)[c]	2.45 (0.70-4.20)[d]	0.61	−3.49 (0.89)	2.99 (0.70-5.29)[c]	3.02 (0.87-5.17)[d]	0.60
Training group (completed homework[e]), n=28	−2.60 (0.78)	2.53 (0.46-4.60)[c]	2.60 (0.67-4.53)[d]	0.63	−4.02 (0.93)	3.52 (1.13-5.90)[d]	3.60 (1.42-5.78)[d]	0.74
Training group (did not complete homework), n=8	−2.14 (1.25)	2.07 (−0.65 to 4.80)	1.93 (−0.58 to 4.45)	0.53	−1.67 (2.25)	1.16 (−3.44 to 5.77)	1.00 (−3.27 to 5.28)	0.23
Control group, n=37	−0.07 (0.65)	-	-	-	−0.50 (0.77)	-	-	-

[a]Adjusted for sex, age, and baseline K6 scores.

[b]Unadjusted Cohen's *d* compared to control group.

[c]*P*<.05.

[d]*P*<.01.

[e]Did homework using I-CBT more than once (mean implementation times was 1.5).

Discussion

Principal Findings

The results of this study suggest that a brief stress management program combining group CBT training and Web-based CBT homework does not provide significant alleviation of stress when analyzed across all participants but does provide moderate alleviation of symptoms in employees with clinically significant psychological distress. Our results further suggest that this type of educational program can improve self-confidence in the ability to cope with stress.

In previous studies of CBT for psychiatric patients, considerable time was required for face-to-face interaction between the CBT expert and patients, and intervention was discontinued in 30% to 50% of cases [24,39,40]. This conventional method is therefore generally considered unfeasible in a workplace setting, as most employees have insufficient time to complete high-intensity CBT and are considered less motivated regarding participation in CBT than psychiatric patients in a clinical setting.

Recently, low-intensity CBT programs have become increasingly popular, and such programs have been implemented to provide medical and psychological support to as many people experiencing psychological distress as possible [21-23]. The positive effects of low-intensity CBT on the mental health of patients have already been reported for group education [41] as well as for therapy via email, phone [24,25], and Internet [26-28]. Further, the discontinuation rate for low-intensity CBT is lower than that for face-to-face CBT [24,42-44].

Van der Klink et al reviewed 49 studies to compare the effects of cognitive-behavioral, multimodal, relaxation, and organization-focused intervention programs [9] on work-related stress. Improvement in psychological distress was greatest for cognitive-behavioral intervention programs such as CBT (d=0.68), followed by multimodal programs (d=0.51), relaxation programs (d=0.35), and organization-focused programs (d=0.08). While the intensity of CBT reviewed by Van der Klink et al was lower than conventional CBT, it is probably still unsuitable for many workplaces as a high degree of care, time, and involvement by experts is required. Of note, in Van der Klink et al's review, the mean number of CBT sessions was 7.6 [9]. Mohr et al reported that face-to-face CBT sessions and CBT sessions via telephone were conducted a total of 16 times, with a mean session time of approximately 45 minutes in their study [24]. The protocol for Web-based CBT by Titov et al required six sessions across 8 weeks, and experts held these sessions on a one-on-one basis [45]. Similarly, in a study of T-CBT by Furukawa et al, 30- to 45-minute sessions were held a total of eight times, and experts (such as therapists) were available for each participant, although no one-on-one sessions were required [46].

The training program implemented in the present study was developed by combining a group CBT course with short-term Web-based CBT. To enable implementation in a workplace setting, the program was simplified even more than conventional low-intensity CBT. Concern of benefits being compromised due to oversimplification is therefore justified. Indeed, although a brief Web-based psychoeducational program was found to have slightly improved the degree of occupational satisfaction, it did not clearly improve self-efficacy or problem-solving skills [47]. To counteract any detractions due to the simplicity of our program, we increased efficacy by asking participants to complete individual homework assignments.

In the present study, analysis across all participants demonstrated alleviation of stress without statistical significance. Two possible reasons for this lack of a substantial finding are that some participants may not have clearly understood the requirements for training, subsequently losing motivation to participate in the CBT program. However, despite the lack of any notable effect of our educational program across all participants, we found that our program did significantly alleviate the stress among those employees who had clinically significant psychological distress before the program. This positive effect was particularly high in the training subgroup that also completed Web-based CBT homework, likely due to the stronger motivation of employees with higher stress to participate and actively complete the homework, thereby alleviating stress. Participants who were provided information concerning CBT during the Web-based CBT are reportedly more likely to complete their homework [48,49]. During the present study, participants were provided CBT-related information four times during the Web program, which might have enhanced their understanding of CBT and thereby increased the positive effects of CBT among those who completed the Web-based CBT homework.

Analysis across all participants shows that the ability of participants to recognize dysfunctional thinking habits increased significantly both immediately after training and on 6-month follow-up. However, these differences disappeared after adjusting for baseline data. The ability to correct dysfunctional thinking patterns between two groups was only significantly different on 6-month follow-up after adjustment. The poor robustness of these results might be due to the lack of statistical power. We are therefore unable to conclude whether or not the ability to recognize dysfunctional thinking habits and correct dysfunctional thinking patterns improved in the present study. Nevertheless, the ability to cope with stress significantly improved on 6-month follow-up in both unadjusted and adjusted analyses. These results suggest that this program improves confidence in stress management skills. Using CBT to teach this new stress management skill to participants might improve various health outcomes [50-52].

Regarding problem-solving skills, participants might have been unable to develop these skills due to the relatively short period of time allocated during group education and lack of Web-based CBT homework on the subject. Lecture sessions (with group work) and homework assignments therefore appear necessary to help participants acquire these problem-solving skills.

Limitations

Several limitations to the present study may prevent our findings from being fully generalizable. First, participants were employees of an information technology company and might therefore have been more likely to consent to Web-based CBT

than the general population. Second, the reliability and validity of the original question items have not been evaluated. Therefore, the results of secondary outcomes cannot be confirmed. Third, this study was non-blinded and the participants in the training and control groups worked in the same office and might have shared information. Fourth, we evaluated one primary outcome and four secondary outcomes, which might have increased the possibility of a Type I error. Finally, the sample size was insufficient because we calculated sample size necessary to obtain an effect size of 0.42. Further, the target population contained a significant number of distressed subjects and participation rate was lower than originally expected, which could have affected the robustness of statistical results.

Validation of the effects of this brief training program on the alleviation of distress and development of stress management in the workplace will require randomized clinical trials in a variety of workplaces with diverse corporate structures.

Conclusions

The results of this study suggest that a brief stress management program combining group CBT and a Web-based CBT can have positive effects on the alleviation of symptoms in employees with clinically significant psychological distress and development of confidence to cope with stress. These brief educational programs based on CBT principles might be an effective preventive measure for addressing current concerns of presenteeism and absenteeism among employees with high levels of stress.

Conflicts of Interest

None declared.

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Abbreviations

CBT: cognitive behavioral therapy
ITT: intention-to-treat
K6: Kessler-6 scale
MI: multiple imputation

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Original Paper

An Interactive Health Communication Application for Supporting Parents Managing Childhood Long-Term Conditions: Outcomes of a Randomized Controlled Feasibility Trial

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Abstract

Background: Families living with chronic or long-term conditions such as chronic kidney disease (CKD), stages 3-5, face multiple challenges and respond to these challenges in various ways. Some families adapt well while others struggle, and family response to a condition is closely related to outcome. With families and professionals, we developed a novel condition-specific interactive health communication app to improve parents' management ability—the online parent information and support (OPIS) program. OPIS consists of a comprehensive mix of clinical caregiving and psychosocial information and support.

Objective: The purpose of this study was to (1) assess feasibility of a future full-scale randomized controlled trial (RCT) of OPIS in terms of recruitment and retention, data collection procedures, and psychometric performance of the study measures in the target population, and (2) investigate trends in change in outcome measures in a small-scale RCT in parents of children with CKD stages 3-5.

Methods: Parents were recruited from a pediatric nephrology clinic and randomly assigned to one of two treatment groups: usual support for home-based clinical caregiving (control) or usual support plus password-protected access to OPIS for 20 weeks (intervention). Both groups completed study measures at study entry and exit. We assessed feasibility descriptively in terms of recruitment and retention rates overall; assessed recruitment, retention, and uptake of the intervention between groups; and compared family condition management, empowerment to deliver care, and fathers' involvement between groups.

Results: We recruited 55 parents of 39 children (42% of eligible families). Of those, about three-quarters of intervention group parents (19/26, 73%) and control group parents (22/29, 76%) were retained through completion of 20-week data collection. The overall retention rate was 41/55 (75%). The 41 parents completing the trial were asked to respond to the same 10 questionnaire scales at both baseline and 20 weeks later; 10 scores were missing at baseline and nine were missing at 20 weeks. Site user statistics provided evidence that all intervention group parents accessed OPIS. Analysis found that intervention group parents showed a greater improvement in perceived competence to manage their child's condition compared to control group parents: adjusted mean Family Management Measure (FaMM) Condition Management Ability Scale intervention group 44.5 versus control group 41.9, difference 2.6, 95% CI -1.6 to 6.7. Differences between the groups in the FaMM Family Life Difficulty Scale

(39.9 vs 36.3, difference 3.7, 95% CI -4.9 to 12.2) appeared to agree with a qualitative observation that OPIS helped parents achieve understanding and maintain awareness of the impact of their child's condition.

Conclusions: A full-scale RCT of the effectiveness of OPIS is feasible. OPIS has the potential to beneficially affect self-reported outcomes, including parents' perceived competence to manage home-based clinical care for children with CKD stage 3-5. Our design and methodology can be transferred to the management of other childhood conditions.

Trial Registration: International Standard Randomized Controlled Trial Number (ISRCTN): 84283190; http://www.controlled-trials.com/ISRCTN84283190 (Archived by WebCite at http://www.webcitation.org/6TuPdrXTF).

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KEYWORDS

child; chronic condition; chronic kidney disease, CKD; family; feasibility; interactive health communication application; online; long-term condition; parent; randomized controlled trial

Introduction

Children and young people (children) aged 0-19 with conditions such as chronic kidney disease (CKD), stages 3-5, often require treatments at home, which can be complex and intrusive. Research into long-term or chronic childhood (hereafter referred to as chronic) conditions helps us understand how families manage the child's condition at home with remote support from multidisciplinary teams (MDTs) [1-5]. CKD, a complex set of disorders with a wide range of primary causes and complications has an unpredictable course. Parents' home-based clinical management responsibilities include fluctuating levels of monitoring and intervention, which can be complicated, intrusive, and require skilled work by families. This skilled work can present extensive challenges for parents and be difficult for them to maintain, especially if they do not possess the comprehension needed to understand instructions. For example, parents may need to collect or test urine samples; understand clinically indicated investigations such as laboratory and imaging studies; administer medications; conduct gastrostomy or naso-gastric tube feeds; set up and run peritoneal dialysis; carefully monitor diet and fluids; and recognize and act on subtle but significant clinical changes in their child. Additionally, parents need to communicate effectively with staff and coordinate many aspects of personal and clinical care while supporting their child, promoting child development, and maintaining normal family life. Parents' failure to become competent at clinical management could lead to non-adherence to treatment regimens, inability to recognize and respond to significant clinical changes, and negative clinical outcomes for the child such as undetected urinary tract infection, which can further damage the kidneys and impair kidney function [2]. In many children, CKD progresses from stage 3 when they require careful monitoring and occasional medications or clinical investigations, to stage 5 when they require renal replacement therapies such as dialysis or transplantation [6] (Table 1).

Table 1. Chronic kidney disease stages 3-5 (adapted from the Renal Association).

CKD stage (GFR[a])	Clinical criteria	Clinical management
3a (35-59%)	Moderately reduced kidney function	Observation, control of blood pressure, and risk factors for progression to stage 4
3b (30-34%)		
4 (15-29%)	Severely reduced kidney function	Planning for end stage renal failure
5 (<15% or on dialysis)	Very severely reduced kidney function or end-stage renal failure	Treatment choices (renal replacement therapies)

[a]GFR=estimated Glomular Filtration Rate.

Families respond in various ways to chronic conditions. Some adapt well to clinical management responsibilities and are able to develop a sense of control over their lives while others struggle to do so. Family response to chronic conditions is closely related to children's clinical outcomes, and non-adherence to prescribed treatments is the primary cause of treatment failure in conditions such as CKD [7-9]. The burden of condition management generally lies with parents and other caregivers rather than the child. A recent systematic review of qualitative studies of parents' views on treatment non-adherence in various medical conditions found caregivers worked hard to retain a sense of control by dealing with challenges such as the child's resistance to treatments. Nevertheless, strict treatment adherence, which is expected by health professionals, could threaten parents' priorities around preserving family relationships and providing a "normal family life" [10,11].

A sense of control has also been associated with the notion of empowerment in pediatric care [12]. Parents want to be empowered to competently deliver clinical care, to recognize and respond appropriately to changes in the child's condition, and to communicate effectively with health professionals about condition management and to relatives, friends, and teachers about the implications of the condition for the child [1,13-16]. Moreover, professionals wish to empower parents, and strategies to help them do this include promoting equal relationships, critical reflection and advocacy, focusing on strengths, supporting active participation and decision-making, providing information, and developing skills [17].

It is important that children with chronic conditions are cared for in ways that minimize emotional trauma and assist in their recovery, and that such ways of delivering care are investigated to see if they are effective [18,19]. In a recent ethnographic study of interactions between fathers, mothers, and professionals during shared care of CKD, it was observed that over time, professionals developed a shared repertoire of tools and artefacts (such as diagrams, anatomically correct dolls, booklets) to support their communications with parents, and that these tools and artefacts helped some parents and professionals to accomplish common ground [4]. Although both parents are often involved in caring for children with chronic conditions, fathers' views tend to be underrepresented in the health care literature [20]. However, evidence is now emerging about the extent of the fathers' involvement and the value of fathers' involvement for children, families, and fathers themselves [20-22].

In addition, family responses to chronic condition management can be affected by individuals' health literacy skills (ie, the ability to comprehend health information) [23]. Parents are increasingly likely to search the Internet for information and support to supplement the guidance they receive from health professionals. However, few interactive, condition-specific, evidence-based online resources are available. Where online resources do exist, they are often based on myth and hearsay [15], and parents with poor literacy levels may be unable to discriminate between high and low quality information and may not be confident in using the Internet [24,25]. Therefore, rigorously developed and evaluated online resources that meet parents' and professionals' needs and preferences are required.

A Cochrane Review [16] shows the use of interactive health communication applications (IHCAs)—computer-based, usually Web-based, information packages for patients/carers that combine health information with social support, decision support, or behavior change support— has positive effects on users. IHCA users tend to become more knowledgeable and perceive higher levels of social support than non-users. Patients who have access to IHCAs either themselves or via a caregiver/parent might have improved behavioral and clinical outcomes compared to non-users, and IHCAs are more likely than not to have a positive effect on users' management ability and self-efficacy. Therefore, more high-quality studies are recommended to determine the best type of and best way to deliver IHCAs, and to establish how IHCAs affect different groups of people with chronic illness [16].

The current study forms part of a phased-approach to development and evaluation of a complex intervention [26], an IHCA for parents of children with CKD stage 3-5. This study was framed by Bandura's concept of self-efficacy, which provides a basis for understanding personal motivation, well-being, and feelings of personal accomplishment in situations that are cognitively, behaviorally, and emotionally challenging [27]. To help parents develop self-efficacy for managing their child's CKD, we first developed an online parent empowerment model in CKD management [28]. The main sources of information that influence perceptions of self-efficacy (mastery experience, vicarious experience, verbal persuasion or similar sources of social influences, and affective states [27]), when integrated with the two main components of our Online Parent Information and Support (OPIS) app, as required by parents and professionals (clinical care-giving support and psychosocial support for care-giving) resulted in the model (Figure 1).

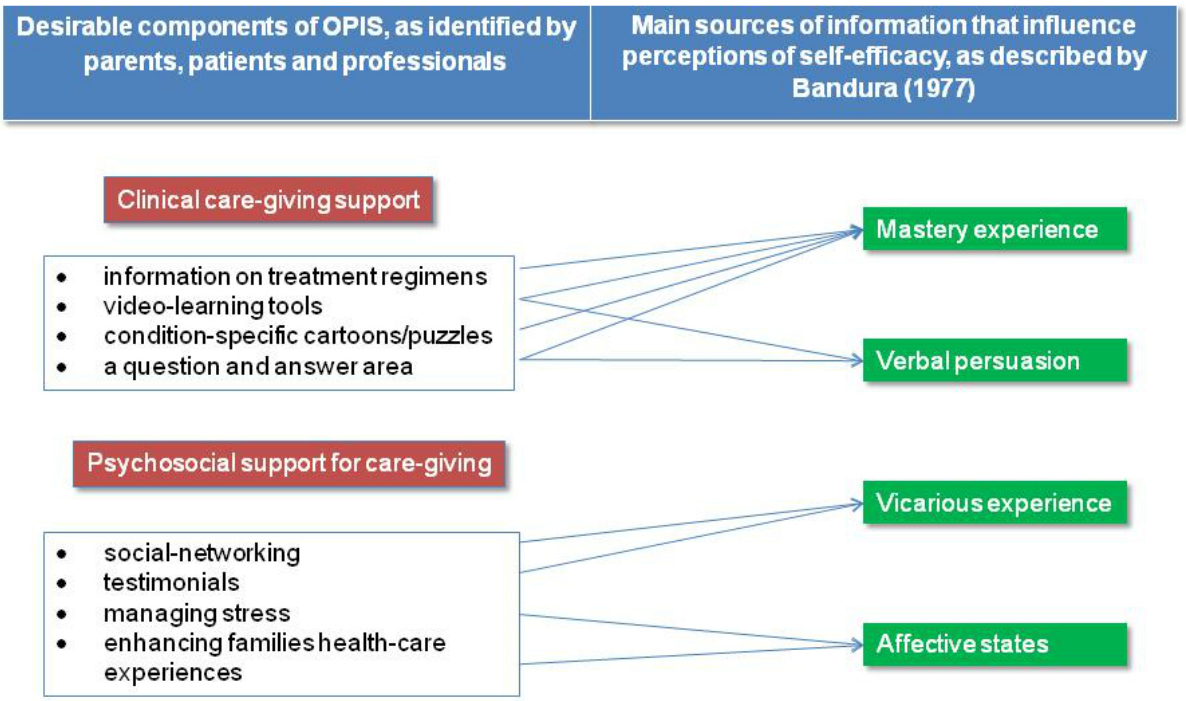
We developed an IHCA, the OPIS application, in collaboration with families and health professionals. The OPIS comprises clinical care-giving support (information on treatment regimens, video-learning tools of MDT professionals explaining how to undertake clinical procedures at home, condition-specific cartoons/puzzles, and a question and answer area) and psychosocial support for caregiving (social networking, testimonials from other parents of children with CKD, and advice on managing stress) [11,28].

We implemented and assessed OPIS for feasibility in the kidney unit of a large children's hospital in the north of England [28,29]. In an earlier report, we provided evidence that parents found OPIS to be very usable and acceptable, and implementation into standard practice was shown to be feasible [30]. In fact, 93% of users reported that OPIS was easy to use and therefore, there was confidence that the design and technology of the ICHA was not a barrier to its use. Qualitative suggestions by parents included refinement of OPIS components, enabling personalization of OPIS functionalities, and proactive endorsements of OPIS by professionals [28].

The purpose of this paper is to build on our previous report [30] by presenting the results of a study that addressed two objectives: (1) to assess the feasibility of a future full-scale randomized controlled trial (RCT) of OPIS in terms of recruitment and retention of mothers and fathers, data collection procedures, and psychometric performance of the study measures in the target population, and (2) to investigate trends in change in outcome measures in a small-scale feasibility RCT in parents of children with CKD stages 3-5.

The results reported here will inform the development and implementation of a future RCT that will be sufficiently powered to detect significant change in outcomes attributable to OPIS. To our knowledge, OPIS is the first IHCA to have been rigorously developed with families living with CKD and health professionals, and then tested with parents.

Figure 1. Online Parent Empowerment Model in CKD management [28].



Methods

Study Design

To achieve the stated objectives, we undertook a small-scale study that used a two-group RCT design with data collected at two points in time: entry to the study at baseline and 20 weeks later (ISRCTN: 84283190). Approval to conduct the study was obtained from the National Health Service (NHS) Research Ethics Committee (REC) (Reference: 11/N/W/0268) and the NHS Trust Research and Development department. No incentives were offered to parents for enrolling in the study.

Treatment Conditions

Parents who provided written informed consent were randomly assigned to one of two treatment conditions: (1) usual support involving discussions with members of the MDT when the child was an in- or out-patient, and for children with stage 5 CKD, home visits from a specialist nurse to teach or reinforce clinical skills, as required (control group), or (2) usual support plus password-protected access to OPIS for 20 weeks, which allowed sufficient time for participants to become familiar with OPIS (intervention group). Children at stage 5 CKD require peritoneal dialysis treatment; the specialist nurse teaches the child and family how to perform this treatment over a series of home visits until they are considered competent to perform it on their own. Future home visits follow to ensure the treatment is being performed correctly. Other clinical skills including giving injections, and managing nasogastric or gastrostomy feeds would also be taught at home with ongoing support to ensure competency.

OPIS was housed on a university Web server and accessible to intervention group parents via their personal computers, mobile phones, tablets, or smartphones. A screenshot of one view of OPIS is shown in Figure 2.

OPIS is Health on the Net (HON) certified (HONConduct443339) [31]. The HON certificate serves as a guarantee that OPIS complies with and pledges to honor the eight principles of the Code of Conduct developed by the HON Foundation: Authority, Complementarity, Confidentiality, Attribution, Justifiability, Transparency, Financial Disclosure, and Advertising. More detailed description and discussion of the development, implementation, usage, and assessment of acceptability and usability of OPIS have been published elsewhere [30,32].

Figure 2. Screenshot of OPIS homepage.

Sample

Parents were considered eligible for this study if their child aged 0-19 years was receiving care at the study site, they had not participated in the development of OPIS and they had access to the Internet via a personal computer or mobile device. Eligible parents were notified of the study by a member of the MDT. Interested parents were referred to the researcher appointed to manage the project and collect/analyze data who then explained study requirements, answered any questions, and obtained written adult consent. As fathers and mothers may have differing information and support needs when their child has a chronic condition [13,20,22,33-36], both parents were invited to participate if they shared the clinical caring role. After baseline data collection, participating parents were then randomly allocated to either the control group or the intervention group at the family level. That is, if two parents of an index child were enrolled in the study, both parents were allocated to the same treatment condition. One of the authors who was not involved in data collection, generated the randomized allocation sequence using nQuery Advisor 6.0, with blocks of random length in an allocation ratio of 1:1, randomized and stratified by CKD stage (3 vs 4/5) and ethnicity (white/black vs South Asian) of the child. Clinical care needs can vary according to CKD stage (Figure 1), and CKD prevalence is higher in UK South Asian groups relative to their representativeness in the overall UK population [37]; thus, we aimed to distribute disease stage and parent race/ethnicity equally between the two treatment groups despite the small scale of the study. The allocation sequence was concealed from parents and the researcher using sequentially numbered opaque envelopes containing the allocation code, which were prepared by a person not otherwise involved in the study. However, blinding parents to whether or not they had been assigned to receive the intervention was not possible. While our working benchmark for feasibility was to have 30 parents in each group by end trial for this pilot/feasibility study [38], we acknowledged from the start that

we would be limited by the number of eligible children under care at the study site at that time.

Data Collection Procedure

Data were collected between September 2012 and September 2013. Measurements were conducted before parents were randomized. Those in the intervention group received a username and password to enable OPIS access, and after 20 weeks at which point their access to OPIS ceased. Data collection took place at a time/place convenient to the parents, either in the family home or a quiet area in the hospital; one interview was conducted by telephone at the parent's request [39]. Evidence is emerging of the persuasive practices of some parents to engage their families in research, which underlines the importance of accessing all potential participants directly, and the importance of sensitization to interactions between family members when engaging in research [37]. Based on this emerging evidence, including our own experience of purposefully recruiting mothers, fathers, and children separately to research (eg, [13,33]), in families where both parents participated in the current study, we ensured that they completed the measures independently of each other. That is, they completed the measures in separate rooms and were not allowed to discuss the measures until each had completed their measure.

Study Measures

At baseline, we used an investigator-devised form to collect background data (child age, sex, postal code, CKD stage at study entry/exit; parent age, sex, race/ethnicity, language, educational achievement, socioeconomic status of neighborhood based on postcode, ethnicity, and clinical care experience). At both baseline and 20 weeks, we administered a set of standardized measures in the following order: the Rapid Estimate for Adult Literacy in Medicine (REALM) [40], the Family Management Measure (FaMM) [8], the Service System Subscale of the Family Empowerment Scale (FES) [41], and the Dads Active Disease Support Scale (DADS) [42]. At baseline, background data were collected first followed by administration of the standardized measures in the order stated. At 20 weeks, the standardized measures were re-administered in the order stated. Completion of the study measures took 40-55 minutes overall.

Parent health literacy was measured using the REALM. The purpose of this was to determine if parents were likely to need help with self-administration of the outcome measures. This assessment requires the parent to read aloud a list of 66 generic clinical words (such as "fat" or "impetigo') arranged in increasing order of difficulty. The score is calculated by awarding one point for each correctly pronounced word and nil for each mispronounced or skipped word. A score of 59 or less indicates low health literacy while a score of 60 or more indicates adequate health literacy. The REALM has face, criterion, and construct validity for use as a health literacy screening tool in the United Kingdom [40].

Parent management ability was measured using the FaMM. The FaMM was developed to measure how families manage caring for a chronic condition and the extent to which they incorporate management into family life. The FaMM has 53 items overall, with 45 items for all parents and eight additional items for partnered parents only. Items are scored from 1-5, meaning strongly disagree to strongly agree. There are five summated scales for all parents measuring the dimensions of Child's Daily Life, Condition Management Ability, Condition Management Effort, Family Life Difficulty, and View of Condition Impact as well as a sixth scale only for partnered parents measuring the dimension of Parental Mutuality.

The FaMM Condition Management Ability Scale (12 items) addresses parents' perceptions of their competence at taking care of the child's condition. Because the intent of the OPIS intervention was to enhance parents' ability to manage their child's CKD, we were especially interested in the Condition Management Ability scale of the FaMM. Higher values mean parents view themselves as more capable of managing the condition. Example items that help to illustrate the concept and its domains include (1) "We have some definite ideas about how to help our child live with the condition", and (2) "We have not been able to develop a routine for taking care of our child's condition" [8].

Parent empowerment was measured using the Service System Subscale of the FES that explores parents' relationships with health professionals and parents' level of comfort in asking questions and voicing their opinions [41]. The FES measures caregivers' beliefs and confidence regarding the services the child needs, their initiative in obtaining these services and making sure that the professionals understand and respect their opinions regarding what the child needs, their knowledge and understanding of services, and their positive attitudes about their ability to obtain and claim the services the child needs. The items are scored in the same direction and higher scores indicate relatively more empowerment in a specific area. The FES has robust psychometric properties and therefore has value in assessing the empowerment status of families.

Father support for managing the child's CKD was measured by the DADS, a 24-item Likert-type scale with separate forms for mothers and fathers. The DADS was developed to assess the support offered by fathers, and mothers' perceptions of the quality of that support. The results of confirmatory factor analysis provide support for the construct validity of the DADS, and two factors (amount and helpfulness of fathers' involvement) best accounted for participants' responses [42]. The DADS has been used in other studies of family response to childhood chronic conditions and the level of reliability was acceptable [21]. To our knowledge, our study represents the first use of the DADS, the FaMM, and the FES, all of which were developed in North America, with parents in the United Kingdom. However, we did not observe any difficulties when parents were completing these measures about the interpretation and understanding of items and response ratings.

Analysis

Data were analyzed using IBM SPSS Statistics Version 20. Participants who dropped out were not contacted further in keeping with REC approval, and the data they provided were compared with those of participants who completed the study. Scores on the outcome measures were calculated and missing values on items handled according to the methods prescribed

by the developers. Consistent with the nature of the study and small sample size, our post-intervention analyses should be interpreted conservatively. Intraclass correlation coefficients (ICC) were estimated for each outcome measure to assess the level of within-family variation, and model performance was measured by estimating the square of Pearson's correlation between actual and predicted values. Confidence intervals were estimated for recruitment and retention rates. The internal consistency of all outcome measures was estimated using Cronbach alpha at baseline across the two groups combined for those completing the study, as the outcomes were measured on a population that had not been previously assessed.

Analysis of trends in change in outcome measures from 20 weeks was adjusted for baseline scores using linear mixed models to allow for having more than one parent participating in a family with adjustments for stratification by CKD stage (3 vs 4/5) and ethnicity (white/black vs South Asian). Confidence intervals and effect sizes for the adjusted differences in means at 20 weeks were estimated—the aim in this feasibility study being to inform sample size estimates for a future full-scale RCT rather than to detect significant differences [38].

Results

Objective 1: Feasibility of Recruitment and Retention and Application Usage

A total of 94 eligible children were identified at baseline. A CONSORT diagram (Figure 3) describes the recruitment and retention of parents through the phases of the feasibility RCT. In total, 39 index children with 55 parents (42% of eligible families invited, 95% CI 32-52) participated. Data at 20 weeks were provided by approximately three-quarters of parents in the control group (22/29, 76%; 95% CI 58-88) and in the intervention group (19/26, 73% of those participating; 95% CI 54-85).

At baseline, the two groups were balanced in terms of the stratification variable ethnicity (white/black/Afro-Caribbean vs South Asian) but not CKD Stage (3 vs 4/5). No parent who recorded a REALM score of less than 60 (ie, low level of health literacy) at baseline withdrew from the study before trial end. Table 2 summarizes parent and child characteristics by group allocation at baseline for those completing the trial.

A quarter of participants (14/55, 25%) were not retained through trial end, including seven from each treatment group. At enrollment, participants were told they could withdraw from the study at any time without providing a reason. However, most parents who withdrew apologized and volunteered a reason (see Figure 2). Control group parents who withdrew tended to be slightly younger than those who were retained (mean ages 37.0 vs 44.1 years); corresponding ages in the OPIS group were similar (41.4 vs 42.7 years). In both treatment groups, parents who withdrew from the study tended to have a lower socioeconomic status based on neighborhood ranking than those who were retained. In the control group, half (5/10) of those whose child had CKD stage 3 were retained through trial end, compared with the majority whose child had CKD stage 4 or 5 (17/19, 89.5%). In the intervention group, the proportions were as follows: child with CKD stage 3 (8/10, 80.0%) versus child with CKD stage 4 or 5 (11/16, 68.6%). Otherwise, characteristics of parents who were retained through trial end were similar to characteristics of those who were not.

Figure 3. CONSORT diagram showing participant flow through study.

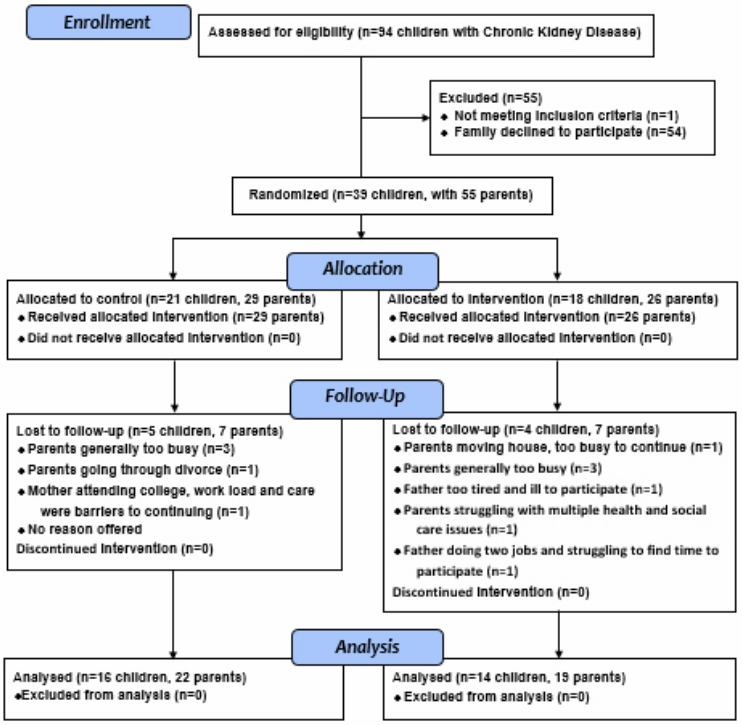


Table 2. Parent and child characteristics by randomization group at baseline for those completing trial.

Characteristic	Control group	Intervention group
Participants, n	22	19
Index children, n	16	14
Parent age, mean (SD)	44.1 (8.3)	42.7 (10.3)
Parent gender, n (%)		
Female	11 (50)	11 (58)
Male	11 (50)	8 (42)
Parent ethnicity, n (%)		
White European	16 (73)	16 (84)
Afro-Caribbean	0 (0)	1 (5)
South Asian	6 (27)	2 (11)
Parent primary language, n (%)		
English	19 (86)	19 (100)
Bengali	2 (9)	0 (0)
Polish	1 (5)	0 (0)
Parent socioeconomic status[a], median (range)	13,041.5 (96-28,036)	21,547 (235-29,472)
Child age in years, mean (SD)	10.2 (5.7)	9.1 (5.5)
Child gender, n (%)		
Female	5 (31)	4 (29)
Male	11 (69)	10 (71)
Child CKD stage, n (%)		
Stage 3	3 (19)	6 (43)
Stage 4	5 (31)	0 (0)
Stage 5	8 (50)	8 (57)

[a]Postal code-based neighborhood ranking (1=highest level of deprivation to 32,482=lowest deprivation).

Application Usage

In total, 19 parents accessed OPIS with a mean of 23.3 visits per user (SD 20.8, range 2-64); OPIS was visited 443 times with a total of 3154 page visits. The mean duration of time spent on the site per visit was 12 minutes, 11 seconds (range 2 seconds to 58.0 minutes). Visits lasted between 10 and 30 minutes, and 88.9% (394/443) of visitors used desktop/laptop computers, 7.9% (35/443) used mobile phones, and 3.2% (14/443) used tablets. Tablet users spent the longest time on OPIS while the desktop/laptop users spent the shortest. The most common depth of visit (25.5%) entailed viewing 20 or more different screens, with parental viewing in 34.6% of these visits ranging from 9-19 different pages. The highest number of page views in 1 week was 541. There were two peaks in usage: one at the start of the study, and the other when users were notified that new research reports regarding CKD were placed in OPIS by the project team. The most popular area was “Kidney Health” followed by “Case Studies” and “What to eat/drink”. The least popular area was “FAQ”. Average time for page download was 1.8 seconds [30].

Qualitative Findings

During parent exit interviews, the issue of usage feasibility was frequently highlighted by parents [30,43], as indicated by the following illustrative quotations. Parents appreciated the opportunity to hear and read accounts on OPIS from other parents of children with CKD: “I think the information was pretty good on it, from the first one when I went onto it, because the main thing is you want to see how other parents are going on with it [CKD management]” [parent/071/mother].

In addition, parents found OPIS easy to access and valued the section that describes the roles and responsibilities of the different MDT members responsible for their child’s overall CKD management: “Because it’s always nice that if you go into somewhere knowing you’ve got a picture of a face with a name, you think ah, yeah, we know her” [parent/056/father].

Parents found the range of information on OPIS interesting: “I enjoyed having a good look around it, I found it interesting for somebody that’s involved in it [CKD management] as a parent” [parent/052/mother].

Furthermore, parents appreciated the links provided to other related websites that had been validated by the OPIS research

team, including MDT professionals: “Really liked trustworthy links page, knowing the MDT has agreed to them, it really helps, knowing it’s the right information and not scaring you half to death!” [parent/045/mother].

The REALM required self-administration for assessment of health literacy. Parents were given the option to self-administer the other study measures or have the researcher read the questions aloud and record the parent’s response. At baseline, 96% (53/55) of the parents opted to self-administer the other measures, and for expediency some chose to do this while waiting for their child’s outpatient appointment. Parents offered two main reasons for preferring self-administration: either (1) they wanted to complete data collection as quickly as possible because of time constraints imposed by their child’s clinical demands and their own personal commitments, or (2) reading the questions themselves helped them to better understand the issues being explored and to consider their response.

Observing while parents self-administered the REALM enabled the researcher to discreetly determine whether a respondent may have difficulty completing the remaining measures without assistance. Most parents had little difficulty completing REALM, but two fathers struggled to read out several words. In these instances, the researcher adopted an encouraging and reassuring tone, explaining that our purpose was not to judge parents but to help us learn from parents how best to explain clinical terms.

Although flexibility and respect for parents’ preferences regarding place and time for data collection is important, the outpatient waiting area was not always appropriate for this purpose. For example, with the two fathers who displayed discomfort with REALM, the researcher suggested a move to a more private location nearby. However, even after the change of location these fathers both read out the words in an increasingly lower register, with a bowed head and constricted body language. In one instance, the researcher stopped REALM administration before it was completed because of the father’s profound reading difficulty and apparent discomfort. The father went on to complete the remaining measures with the researcher reading questions aloud to him.

At baseline, members of the control group had the four lowest REALM scores (10, 42, 43, and 43); a member of the OPIS group had the fifth lowest score (57). The REALM score at both baseline and 20 weeks was recorded for 82% (18/22) of parents in the control group and 95% (18/19) of parents in the intervention group. Of these parents, 86% (31/36) scored 60 or above (denoting an adequate level of health literacy) at both time points. The remaining 5 parents scored less than 60 (denoting inadequate health literacy) at both time points and 4 of these parents were in the control group.

Administering the FaMM (the measure with the next highest number of items) immediately after the REALM meant that the researcher could reassure parents that the remaining measures (FES and DADS) would not be as time consuming to complete. After completing the FaMM, all parents volunteered that it had helped them to appreciate the amount and level of clinical care they provided for their child. They also said the FaMM prompted them to reflect on issues such as their child’s education and well-being, and how they enabled their child to achieve a good quality of life. While completing the FaMM, two mothers became emotionally distressed as they recalled the burden of care management; the researcher offered to suspend the interview if it was becoming burdensome or to arrange a meeting with a member of the MDT for counseling. Both mothers declined this offer citing the therapeutic benefit for them of processing these emotions and that through participating they were making a positive contribution to future management by other families. A few parents (8/55, 15%) parents stated that some items on the FaMM (such as “It seems as if our child’s condition controls our family life” and “Our child’s condition requires frequent hospital stays”) were not appropriate to their current situation.

The Service System subscale of the FES appeared to be easily understood by parents as no clarification was requested. The DADS scale was also easily understood.

Psychometric Performance of the Study Measures in the Target Population

Table 3 shows descriptive statistics and reliability estimates for the study measures across the total sample at baseline for the 41 parents who completed the study. Missing values were few across the 10 measures at baseline and 20 weeks. With regard to the Table 3 column entitled, “percentage relevant scored”, all 41 parents were scored on five of the six FaMM scales at both time points; the 6 parents without a score on the Parental Mutuality Scale were participating in the study as a single-parent family. However, these 6 parents may have had a parenting partner (marital status was not sought), but if there was a partner, then he/she was not a study participant. The DADS scales were also designed for 2-parent families; the same 6 parents plus another 3 and 2 parents respectively had missing values on the DADS Amount or Helpfulness scales, again at both time points. Two parents did not complete the Service System Subscale of the FES at both time points, while 5 parents did not complete the REALM at baseline and 4 did not complete it at 20 weeks.

The majority of the outcomes measures had an acceptable level of internal consistency reliability; a Cronbach alpha ≥.70 is commonly considered as being acceptable in psychosocial research. The two exceptions were the FaMM Condition Management Ability Scale (alpha=.52) and Condition Management Effort Scale (alpha=.62). In the Condition Management Ability Scale, the item “We have enough money to manage our child’s condition” was negatively correlated with eight of the other 11 items, and its correlation with the sum of the other items was -.21. Deleting this item from the scale increased Cronbach alpha from .52 to .62. Another two items, “We have some definite ideas about how to help our child live with the condition” and “We often feel unsure what to do regarding our child’s condition”, showed very little correlation (r<.04) with the sum of the other items, but separately deleting either of these would have raised Cronbach alpha by only .02. We decided to delete the item “We have enough money to manage our child’s condition” from the Condition Management Ability Scale, and the following analyses are based on the revised version of this scale.

Parents had not needed several items in the DADS during the previous 6 months. For example, 20 parents had not attended a support group or educational workshop about their child's condition and had not needed to pay medical bills. Only two parents had complete entries for the Amount score. Table 4 shows descriptive statistics at baseline for the questionnaire scores by randomized group also for parents who completed the study. On the whole, mean scores of most outcome measures in the two groups tended to be similar at baseline.

Table 3. Descriptive statistics and reliability estimates for the study outcome measures at baseline across total sample for parents completing the trial.

Outcome measure	Items, n	Complete responses, n	Number scored, n	Percentage relevant scored, %	Mean (SD, range)	Cronbach alpha[c]
Family Management Measure						
Child's Daily Life Scale	5	39	41	100.0	17.3 (5.0, 10-25)	.72
Condition Management Ability Scale	12	41	41	100.0	45.0 (5.9, 27-56)	.52
Condition Management Ability Scale (revised)[a]	11	41	41	100.0	43.6 (5.4, 32-55)	.62
Condition Management Effort Scale[b]	4	41	41	100.0	14.0 (3.6, 6 -20)	.62
Family Life Difficulty Scale[b]	14	40	41	100.0	36.1 (12.3, 14-56)	.90
Parental Mutuality Scale	8	35	35	100.0	33.4 (6.2,19-40)	.79
View of Condition Impact[b]	10	38	41	100.0	30.1 (6.3,14-41)	.69
Family Empowerment Scale						
Service System Subscale	12	38	39	95.1	4.2 (0.5, 3.1-5)	.85
Dads' Active Disease Support Scale						
Amount score	24	33	33	94.3	79.2 (21.2, 50.1-120.0)	.91
Helpfulness score	24	32	34	97.1	73.0 (19.9, 33.0-112.0)	.95

[a]Excluding the contradictorily correlated item "We have enough money to manage our child's condition".

[b]Higher scores are undesirable.

[c]Based on complete responses for each scale.

Table 4. Outcome scores at baseline by randomized group for parents completing the trial.

Outcome measure	Control group (n=22)			Intervention group (n=19)		
	n	Mean (SD)	Range	n	Mean (SD)	Range
Family Management Measure						
Child's Daily Life Scale	22	17.6 (5.6)	10-25	19	17.0 (4.4)	11-25
Condition Management Ability Scale (revised)	22	42.8 (4.8)	36-52	19	44.5 (6.0)	32-55
Condition Management Effort Scale[a]	22	13.1 (3.9)	6-20	19	14.9 (3.0)	8-20
Family Life Difficulty Scale[a]	22	35.3 (13.8)	14-56	19	37.0 (10.6)	15-56
Parental Mutuality Scale	19	32.8 (6.3)	19-40	16	34.1 (6.1)	20-40
View of Condition Impact[a]	22	30.3 (6.0)	20-41	19	29.8 (6.8)	14-40
Family Empowerment Scale						
Service System Subscale	17	4.3 (0.5)	3.1-5.0	16	4.1 (0.5)	3.2-4.9
Dads' Active Disease Support Scale						
Amount score	17	84.4 (22.5)	50.7-120.0	16	73.9 (18.7)	50.1-115.2
Helpfulness score	18	69.9 (21.1)	41.8-102.0	16	76.6 (18.4)	33.0-112.0

[a]Higher scores are undesirable.

Objective 2: Trends in Change in Outcome Measures

Table 5 presents the estimated marginal means for the outcome measures at 20 weeks by randomized group for parents completing the trial, adjusted for baseline scores, stratification variables, and number of parents participating. The ICCs indicate the proportion of variance that can be attributed to differences between families. High values indicate more variation between families (ie, less variation between parents), while low values indicate less variation between families (ie, more variation between parents). Among those whose child's other parent participated in the study, parent dyads showed most agreement in outcomes concerning the practical impact of their child's condition (FaMM View of Condition Impact and Family Life Difficulty Scales). They showed least agreement on the support given by the other partner or the father (FaMM Parental Mutuality Scale and DADS Helpfulness score). While the numbers of parents involved were small, this finding requires further investigation in a fully powered trial. The most noticeable difference was in the change in DADS Helpfulness in the intervention group; mothers' mean score for their partner increased by 15.0 (n=7) from baseline to end trial while fathers' self-perceived mean score fell by 6.0 (n=5). Corresponding changes in the control group were a decrease of 3.5 (n=6) reported by mothers and a decrease of -0.2 reported by fathers.

Table 5. Estimated marginal means[a] for outcome scores by randomized group and their differences at end trial using linear mixed model.

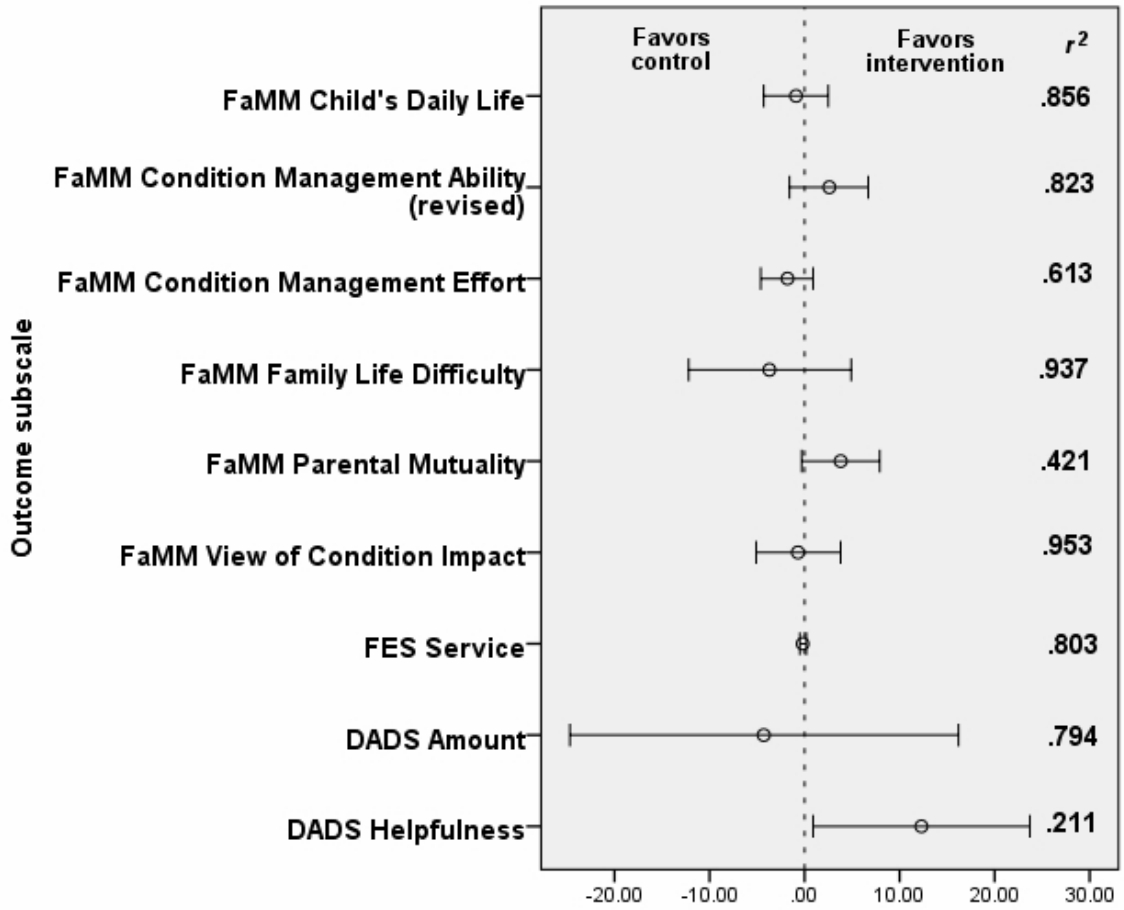
Outcome measure	Control group (n=22)		Intervention group (n=19)		Intervention minus Control			
	n	Mean[a] (95% CI)	n	Mean[a] (95% CI)	Diff (95% CI)	*P* value	r^2	ICC
Family Management Measure								
Child's Daily Life Scale	22	16.9 (13.8-19.6)	19	15.7 (12.8-18.7)	-0.9 (-4.3 to 2.5)	.576	.856	.568
Condition Management Ability Scale (revised)	22	41.9 (38.5-45.4)	19	44.5 (40.9-48.1)	2.6 (-1.6 to 6.7)	.213	.823	.440
Condition Management Effort Scale[b]	22	13.3 (11.0-15.6)	19	15.2 (12.8-17.6)	1.8 (-0.9 to 4.6)	.176	.613	.247
Family Life Difficulty Scale[b]	22	36.3 (29.0-43.5)	19	39.9 (32.5-47.3)	3.7 (-4.9 to 12.2)	.389	.937	.778
Parental Mutuality Scale	19	31.0 (27.7-34.3)	16	34.8 (31.4-38.2)	3.8 (-0.3 to 7.9)	.066	.421	.138
View of Condition Impact[b]	22	29.9 (26.1-33.7)	19	30.6 (26.8-34.4)	0.7 (-3.8 to 5.1)	.763	.953	.829
Family Empowerment Scale								
Service System Subscale	21	4.3 (4.0-4.6)	18	4.2 (3.9-4.5)	-0.2 (-0.5 to 0.2)	.404	.803	.456
Dads' Active Disease Support Scale								
Amount score	17	78.1 (61.3-94.8)	16	73.8 (57.5-90.2)	-4.3 (-24.7 to 16.2)	.667	.794	.614
Helpfulness score	18	70.0 (60.9-79.0)	16	82.3 (72.6-91.9)	12.3 (0.9-23.7)	.036	.211	.161

[a]Estimated marginal mean adjusted for baseline score, severity of chronic kidney disease, ethnicity, and number of parents in family.

[b]Higher scores mean worse outcomes.

After 20 weeks, parents in the intervention group had an adjusted mean score on the FaMM Condition Management Ability Scale (revised) that was 2.7 points better than that for parents in the control group (95% CI -1.6 to 6.7). The linear mixed model for this outcome was a good fit to the patterns in the data (r^2=.823). The findings suggest that parents using OPIS tended to perceive themselves to be managing their child's condition better than parents in the control group perceived themselves to be managing. The adjusted mean for the FaMM Family Life Difficulty Scale was 3.7 points worse in the intervention group (95% CI -4.9 to 12.2, r^2=.937). This suggested that parents using OPIS tended to perceive having more difficulties with family life due to their child's condition than parents in the control group perceived themselves to have. Two outcome measures (the FaMM Parental Mutuality Scale and the DADS) applied to families with two parents participating in the study. For the FaMM Parental Mutuality Scale, the adjusted mean at 20 weeks was 3.8 points better (95% CI -0.3 to 7.9, r^2=.421) for the OPIS group than for the control group. Similarly, the DADS Helpfulness score was 12.3 points better (95% CI 0.9 to 23.7, r^2=.211) in the intervention group than the control group. That is, using OPIS tended to improve parent mutuality as measured by the FaMM and perceived helpfulness as measured by the DADS. While this feasibility study was not powered to detect significant differences, these four between-group differences seem clinically meaningful, and if these tendencies were to have held up as more parents were recruited to the study, the differences would have been statistically significant. Given the small size of the study sample and even smaller sizes of the treatment groups, these results require examination in a larger and fully powered RCT. Figure 4 presents the differences between the study groups in marginal means at end trial in terms of whether the difference between baseline and week 20 scores on the various outcome measures indicated an advantage to the OPIS group or an advantage to the control group

Figure 4. Differences in marginal means between the study groups in terms of whether the difference favored the intervention (OPIS) group or the control group (means adjusted for baseline score, severity of chronic kidney disease, ethnicity, and number of parents in family).



Discussion

Principal Findings

The main findings of this study are that a full-scale RCT of the effectiveness of OPIS is feasible and that OPIS has the potential to beneficially affect self-reported outcomes, including parents' perceived competence to provide home-based clinical care for children with CKD stage 3-5. In this section, we first address objective 1 by assessing feasibility of a future full-scale RCT of OPIS. Then we address objective 2 by considering the trends in change on study outcome measures.

Feasibility of Recruitment, Retention, and Data Collection

The results support the feasibility a full-scale RCT. However, in future research recruitment could be improved by examining the potential influences on recruitment to this study. For example, parents could view participating in a study relating to their child's health care with no promise of benefit to themselves or their child, as adding burden to an already stressful situation. Due to the complex care needs of many children with CKD, in particular those with level 4/5 CKD, the unpredictable nature of individuals' disease progression and the potential for the child's condition to deteriorate during the trial, some parents might have declined to participate because of time requirements and duration of commitment.

The researcher recruiting parents and collecting and analyzing data was not a health professional, so not a member of the MDT. Parents were unlikely to feel obliged to agree to the study when approached by the researcher in the way they might have if recruited by a MDT member. We believe it important that the MDT was seen to endorse the research and the researcher. This happened through our strategy of arranging for a professional to introduce the parents to the study during a clinical consultation. This was important because parents often have a long-standing and trusting relationship with members of the MDT and so may have wished to discuss the study with them before making a decision about participation. In future research, more proactive MDT endorsement of the study, such as by referring to OPIS and/or demonstrating it when providing specific information to parents about the child's condition, could further enhance recruitment.

The study design required that parents be randomly allocated to a treatment group. Some parents who were initially interested in the study could have declined enrollment once they realized there was no certainty of allocation to OPIS. Parents possibly also felt an obligation to express interest in the study to the professional who notified them of the study but later felt able to tell the researcher that they were not interested once they realized that their refusal would not be reported to the MDT. In a future full-scale trial, we can adopt a number of additional strategies to potentially increase recruitment/retention. For example, parents usually have several individual outpatient

consultations with members of the MDT at one appointment. We capitalized on the opportunity presented by this as, once the first professional had notified the parents of the study, the researcher then approached them to explain more about what would be involved. Some parents might have been too distracted by their child's presence and the other pending consultations to be able to give due consideration to the possibility of participating and may have preferred an explanatory telephone call from the researcher at a later date. To enable this, the MDT member who initially informed the parents about the study would also need to ask whether parents would consider either an explanatory phone call from the researcher at a later date or a face-to-face meeting with the researcher in clinic on the same day.

In addition, we could alter the study design so that randomization would be to either the intervention (OPIS) or to a wait-list control group. The wait-list control group could elect to receive the intervention immediately after the post-test assessment [44]. Alternatively, we could release basic OPIS content to both groups (eg, the written, purely informational or technical content) and other OPIS content (eg, the interactive resources to promote clinical caring skills and access to the family-to-family area) to the intervention group only. In addition, data collection, particularly at end of trial may be enhanced as a way to improve retention by offering parents the option to provide data via online means and entirely at their convenience [45].

At the initiation of baseline data collection, the REALM was a useful tool to help the researcher determine whether a parent appeared to struggle with reading. This meant the researcher could alter the data collection strategy accordingly to minimize embarrassment for parents with low health literacy and maximize the quality of the data. The measurements of health literacy were stable from baseline to end of trial, which is understandable since health literacy is a relatively stable construct and OPIS was not intended to improve health literacy. The REALM proved to be difficult for 2 parents due to its large number (66) of items. A recently validated Rapid Estimate of Adult Literacy-Short Form (REALM-SF) comprising only seven items could be more appropriate for a future full-scale RCT of OPIS [46]. However, being asked to complete a health literacy assessment tool for a research project can be embarrassing for parents no matter how sensitively it is delivered. One study found that 40% of patients with low literacy felt ashamed about this [47]. Parents with low health literacy who are responsible for reading and understanding complex information and instructions on how to manage their child's condition may feel ashamed and also concerned that they might not be able to safely deliver their child's clinical care. At the same time, they may be embarrassed to tell the MDT that they have health literacy problems in case the professionals would consider them incompetent to enact clinical caring. This fear could have caused great worry for parents who struggled with the REALM. Using the shortened form in future studies would still elicit valuable information while minimizing parental embarrassment and worry. To reduce these problems in the future full-scale RCT, we will administer the REALM-SF at baseline only.

Some potential reasons for attrition in the intervention group are that (1) we adopted a non-directive strategy in that once intervention group parents had received the password and login advice, and (2) we did not direct parents' use of OPIS as we wished to determine parents' undirected usage. Parents might have expected more direct and continued engagement with the study team rather than self-directed exploration of OPIS. In addition, parents might have expected more endorsement by MDT members than was possible within the study resources; indeed our qualitative interviews with parents at study exit confirm this [30]. Parents were most likely to have accessed OPIS resources that seemed particularly relevant to their child's situation and their own health literacy level. Encountering information that provoked uncertainty, fear, and anxiety about their child's future could then have led to their attrition from the study to avoid further exposure to upsetting information. Furthermore, those who remained in the study through to trial end could have limited their exploration of OPIS, which in turn might have attenuated OPIS effects on study outcomes. In a future study, we will adopt a more engaged and directive approach with participants as a potential means to control attrition and improve retention. For example, we are considering using a combination of targeted delivery of the intervention at regular points in the study period using baseline measurements of personal characteristics and preferences to tailor level of detail and content on an individual basis, at particular time points. We would also interview parents to find out which parts of the OPIS website they found to be most and least relevant.

Retention in research that involves Internet interventions has been identified as a major problem and is now widely recognized as a science of attrition [48]. This body of work has a focus on understanding how the reach of the Internet might increase enrollment of patients at greater risk of attrition, incorporating components into trial design to prevent this effect through understanding patient characteristics associated with higher rates of attrition. While initial attempts to find solutions to the problem of attrition may benefit from investigator intuition and trial-and-error approaches, the area of Internet intervention has advanced to a stage where this delivery modality could benefit from more refined and better specified models, which define the components of individual characteristics, human interaction, and person-to-person support that seem to contribute to adherence. Alternatively, for the intervention group in our study, it may be that some parents did not have time to participate in the trial; they might have been struggling with the clinical care demands meaning that accessing OPIS was an additional demand on their time.

While the numbers dropping out of our study for the different measures were between 4 and 7 parents, patterns of attrition tended to differ between the treatment groups. Although the attrition pattern in the intervention group was inconsistent, those dropping out tended to perceive more problems with family life and more impact caused by their child's condition at baseline than those intervention group parents who were retained in the study. This finding is consistent with the finding that the scores on these measures were "poorer" in the intervention group than in the control group (see Table 2). This would need to be monitored in a larger study.

Trends in Change on Study Outcome Measures

To address objective 2, we investigated trends in change on outcomes in a small-scale preliminary RCT in parents of children with CKD stages 3-5. The results suggest that OPIS could improve parents' ability to manage their child's condition more than standard care over 20 weeks. For example, after accessing OPIS for 20 weeks parents were less likely to endorse the statement "We have not been able to develop a routine for taking care of our child's condition". When children have a chronic condition such as CKD, parents usually assume the roles of care coordinator, clinical expert, and advocate as well as their normal parenting roles. Health care providers are uniquely positioned to assist parents in meeting those challenges, and researchers recommend that they aim to promote parents' competence and confidence in their child's care through understanding common challenges that parents face, promoting parent-to-parent connections, and building partnerships with parents and their children with clinical needs [49]. Our trial indicates that OPIS appears to address these recommendations. Although the numbers are very small, there seems to be a suggestion that staying in the OPIS group was harder for South Asians, and dropping out from the control group was more common among parents with a child with CKD stage 3 who may be less needy for support and information. These factors may have implications for setting up a full trial, and such attrition would have to be monitored closely. In a full-scale RCT, we would add a measure of self-efficacy, which is the construct that frames the study [50].

The FaMM Parental Mutuality Scale indicated that parents in the intervention group tended to show less decline in satisfaction in working together over time. Reasons for decline in satisfaction should be explored in a larger study. A smaller drop in parental mutuality in intervention group scores on this measure concurred with more desirable change in the DADS Helpfulness scale. A strong correlation between the FaMM Parental Mutuality Scale and the DADS Helpfulness (correlation) subscale suggests that it would not be necessary to include both the FaMM and the DADS in our future full-scale RCT, thus reducing participant burden.

The FaMM View of Condition Impact Scale showed that both groups' perceived seriousness of the child's condition tended to show improvement, and more so in the control group as compared to the intervention group.

The FaMM Family Life Difficulty Scale showed that the control group parents appeared to perceive fewer difficulties after 20 weeks, but the intervention group appeared to perceive slightly more. It was not clear whether this was a negative or a positive impact, since OPIS may have elevated parents' awareness of their caregiving responsibilities. This would need investigating in a larger study using a measure of anxiety as there could be implications for clinical practice.

We also know that adherence to medical recommendations deteriorates over time [51], which has implications for length of participant involvement in the future RCT. Allowing access to OPIS for a longer time period and/or "booster" doses of OPIS to maintain the durability of desired effects are some options to be considered for future studies.

Information management, specifically limiting awareness of information that generates uncertainty, fear, and anxiety has been well established as a typical parental management strategy in the context of life-threatening chronic childhood conditions [52]. However, parents, in particular low-income parents, may be unable to distinguish between high- and low-quality information and may not be confident in using the Internet [24]. This suggests that IHCAs developed between professionals and families and endorsed by professionals for day-to-day use are more likely to reduce anxiety than to increase it. In future research, we could assess parents' uncertainty tolerance, anxiety, and coping strategies pre- and post OPIS. We could also apply psycho-immunization strategies. Psycho-immunization involves exposing individuals first to relatively benign or generic information and then to increasingly more threatening or relevant information, gradually building up tolerance to uncertainty and controlling the intensity of anxiety and other emotional responses [53].

IHCAs such as OPIS might have the potential to increase fathers' involvement in disease management if suggestions for refinement and usability issues that we reported elsewhere [54] are addressed in future OPIS development and testing. The DADS Amount scores demonstrate that fathers appreciably contributed to their child's care; in fact, two participating fathers administered the majority of clinical care because the mother lacked confidence in managing the skills required. The OPIS prototype seems to support and promote collaborative clinical caregiving by fathers and mothers.

Limitations

A limitation of this study is the low number of participating parents of South Asian descent, despite the relatively high prevalence of CKD in their children. In addition to reasons given by these parents for declining to participate (eg, no time or child transferring to adult care), cultural and language barriers to participation might have been in play. South Asian women, who are often the primary caregivers, can possibly lack knowledge of health risks, have ideas about self-care that differ from those held by white women of European descent, experience language barriers, be subject to the stress of emigration and isolation, be preoccupied with their family's needs, and not seek access to health promotion programs [55]. Our study was a small, non-powered pilot study, so it could not be all-inclusive given its size, and it was not our intention that the results would be generalizable. Given that children of South Asian descent experience heightened risk for CKD and also parental difficulty in managing CKD, strategies to recruit and retain South Asian parents must be addressed. We did involve South Asian parents in in the Study Steering Group, but their involvement in designing the recruitment strategy will strengthen this aspect of our future research.

The number of eligible children being cared for at the site during the study period was less than anticipated, and we did not achieve the target of data from 30 parents per group at trial end. The future full-scale RCT could recruit parents from multiple sites, with the intervention being delivered via the Web as in the feasibility study. Overall, the data obtained from our 51 participants were highly informative.

Conclusion

Our results indicate that a full-scale RCT of OPIS is feasible. Furthermore, being in the intervention group improved parents' perceived management ability to a greater extent than usual care over 20 weeks. Specifically, the FaMM Condition Management Ability Scale appeared to show beneficial change when reinforced by accessing OPIS. A full-scale trial of OPIS is indicated that would include a shorter measurement of health literacy and additional measurements of self-efficacy and anxiety, with an embedded qualitative component to investigate reasons underlying changes in outcome scores.

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Conflicts of Interest

None declared.

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Abbreviations

CKD: chronic kidney disease
DADS: Dads Active Disease Support scale
FaMM: Family Management Measure
FES: Family Empowerment Scale
HON: Health on the Net
IHCA: interactive health communication application
MDT: multidisciplinary team
NHS: National Health Service
OPIS: Online Parent Information and Support
REALM: Rapid Estimate of Health Literacy
REC: Research Ethics Committee

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Proposal

Development of an Evidence-Based Clinical Algorithm for Practice in Hypotonia Assessment: A Proposal

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Abstract

Background: Assessing muscle tone in children is essential during the neurological assessment and is often essential in ensuring a more accurate diagnosis for appropriate management. While there have been advances in child neurology, there remains much contention around the subjectivity of the clinical assessment of hypotonia, which is often the first step in the diagnostic process.

Objective: In response to this challenge, the objective of the study is to develop and validate a prototype of a decision making process in the form of a clinical algorithm that will guide clinicians during this assessment process.

Methods: Design research within a pragmatic stance will be employed in this study. Multi-phase stages of assessment, prototyping and evaluation will occur. These will include processes that include a systematic review, processes of reflection and action as well as validation methods. Given the mixed methods nature of this study, use of NVIVO or ATLAS-ti will be used in the analysis of qualitative data and SPSS for quantitative data.

Results: Initial results from the systematic review revealed a paucity of scientific literature that documented the objective assessment of hypotonia in children. The review identified the need for more studies with greater methodological rigor in order to determine best practice with respect to the methods used in the assessment of low muscle tone in the paediatric population.

Conclusions: It is envisaged that this proposal will contribute to a more accurate clinical diagnosis of children with low muscle tone in the absence of a gold standard. We anticipate that the use of this tool will ultimately assist clinicians towards moving to evidenced based practice whilst upholding best practice in the care of children with hypotonia.

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KEYWORDS

hypotonia; clinical assessment; low muscle tone; evidenced-based

Introduction

In recent years, the importance of early intervention and developmental programs has gained momentum and increased attention in order to promote general child health and well-being [1]. However, in order for early intervention to be optimally effective, early detection is paramount. As clinicians become progressively more involved in early examination, evaluation, and early intervention programs, the accuracy of these early examinations becomes essential in order to contribute to cogent decisions for intervention [2]. Accurate assessment and diagnosis is also essential in order to predict clinical course, manifestations, complications, prognosis, and to provide parental counselling.

Muscle tone assessment is an essential component during the neurological examination of infants and children. It is often crucial to the establishment of an accurate diagnosis and appropriate management of a child. A challenge for clinicians

however, remains in the accurate identification and quantification of muscle tone due to the subjective nature of the clinical evaluation process [3]. This continues to confuse clinicians, and the scientific community has yet to gain consensus on the operational definition, the diagnostic criteria used to determine hypotonia, and the clinical assessment techniques for evaluation [4,5].

Hypotonia can be a confusing clinical presentation, often leading to inaccurate evaluation and unnecessary investigations due to the subjectivity of the assessment [6,7], which is frequently indicated in the literature [3-5,8-11]. If this confusion and uncertainty continues, it may have serious diagnostic implications (eg, psychological repercussions for both the child and family in terms of false labelling, invasive neurological investigations etc). Moreover, clinicians will continue to face the difficulty of accurately and consistently identifying children with hypotonia and diagnosis of the underlying cause. This may lead to a series of negative consequences with respect to proper management of these children and may also pose difficulty in conducting further research on the efficacy of intervention strategies or in the comparison of studies and an inability to apply or generalize results of studies to individuals.

There is evidence in the literature to indicate that the use of care pathways, algorithms, and practice guidelines, in clinical research will help in standardizing care and provide the necessary requirements for effective diagnostic and counselling interventions [12]. These guidelines and tools may also assist in providing a standard flowchart of the evidence-based diagnostic and treatment to be provided for a spectrum of diseases and disorders [12].

Although no gold standard currently exists, some researchers have begun to address the issue of criteria that may be used to assist in assessment of low muscle tone [4,5]. Thus, in order address the current challenges in clinical assessment of hypotonia, and in the move towards more evidence-based assessments, we will develop and validate a clinical algorithm for the assessment of hypotonia in children. The purpose of this tool would be to assist clinicians (paediatricians, occupational therapists, physiotherapists) in the decision making process in a stance towards more objective and accurate clinical diagnosis and making the appropriate referrals for early intervention. Such a study will also speak to the need for more evidence-based assessment and interventions as well as address the global health needs with respect to early detection and intervention. This will inevitably and indirectly advance the attainment of goal four, which is to reduce child mortality, of the United Nations Millennium Development goals [13].

Methods

Overview

This design research process follows the pragmatist paradigm. Above paradigmatic purism, the most appropriate approaches to the study are cross-stage mixed-methods. A three-phase approach will be used in this study.

Phase 1 (Preliminary Phase): Identifying the Gaps and Analysis of the Problem

Phase 1 of the study will assist in the identification and analysis of the problem. We aim to undertake a systematic review to identify and appraise existing assessments that can be used by clinicians (occupational therapists, physiotherapists, and paediatricians) to detect hypotonicity in children. This will also assist in the identification of gaps that will further guide this research process.

Additional work indicated the need for more objective measures in the assessment of hypotonia [14]. A further study [15] gained consensus on the criteria, tests, and methods that would be most appropriate in determining whether a child has low muscle tone. These studies will be used in addition to the systematic review for phase 2 of the study.

Phase 2 (Prototyping Phase): Development of the Clinical Algorithm

Overview

This phase of the study will involve the development and refinement of the clinical algorithm for the assessment of low muscle tone that is guided by theory, existing principles, and technology.

Instrument Development

The desktop approach in this phase will employ inductive, abductive, and deductive logic to the data from phase 1. This will develop from reflection upon experience (interpretivist), as well as from logical speculation from empirical data (positivist), in order to contribute towards the development of the clinical algorithm.

This process involves superimposition of the International Classification of Functioning (ICF) within the algorithm. The ICF also has merit in that it is a universal model that is holistic, based on human functioning not merely disability and is integrative, not merely medical or social. It is also operational and not driven by theory alone [16].

In order to ensure good practice for development of the algorithms, guidelines on the formal development of an algorithm will be considered. Formulation of evidence-based algorithms are said to be an increasing practice in both scientific papers and text books, however, their usefulness is questioned, as many of the authors are found not to adhere to the formal requirements [17,18]. Algorithms are said to be correct if formulated in accordance with technical regulations by the International Organization for Standardization (ISO) norm where each of the different symbols graphically represent the sequence for the solution to a problem [19]. The International Telecommunication Union (ITU-T) norm symbols that have been based on the ISO norms, have been incorporated approximately over a decade ago to adapt the algorithm for clinical practicability [17,19]. These will be considered in the technical aspects in the development of the algorithm in this study.

Instrument Refinement

This phase of the study will involve the refinement of the clinical algorithm for the assessment of low muscle tone with the interplay between reflection and action processes. Clinicians, opinion leaders, and clinical researchers who meet specified inclusion criteria will be solicited during conference workshop sessions as well as structured critique groups in order to implore feedback on the algorithm. We will employ nonprobability purposive sampling. The three clinical disciplines included in this study, will be considered a homogenous group as they belong to the same subculture or have similar characteristics [20]. Individuals will not be selected in an attempt to represent the general population, but rather in their ability to expertly contribute to the research process [21]. An iterative cycling process between phases of reflection and action will occur. Observation and recording of the process and outcomes of the workshop and structured sessions will occur, followed by reflection and refinement through process of modification, reframing, and rejection if necessary.

Phase 3: Assessment and Validation

The methodological rigor with which the algorithm was developed will be subject to the critique of identified experts in the field with the use of the the Appraisal of Guidelines for Research & Evaluation II (AGREE-II) instrument developed by the AGREE Research Trust. The instrument is a generic tool designed primarily to help guideline developers and users assess the methodological quality of guidelines [22]. This tool consists of 23 key items organized within six domains followed by two global rating items. Each domain captures a unique dimension of guideline quality. All items are rated on a 7-point Likert scale. A quality score is calculated for each of the six AGREE-II domains. This will help access the rigour with which the algorithm was developed.

Additionally, we aim to establish epistemic correlation and content validity of the algorithm, via nonprobability purposive sampling of experts, which will include *inter alia*, international leaders in the field of paediatrics as highlighted by authorship in related articles identified in phase 1 (systematic review). A survey consisting of both open and closed ended questions will be developed after phase 2 of the study. Qualitative software packages such as NVIVO or Archiv fuer Technik, Lebenswelt und Alltagssprache-text interpretation (ATLAS-ti) will be used to code qualitative data and the Statistical Package for the Social Sciences (SPSS) version 21 will be used to quantify the responses on the survey. Both content analysis and descriptive statistics will be utilized to analyse the data. Further decisions related to analysis will be determined following the design of the questionnaire. If levels of agreement are necessary, Cronbach alpha will be computed and *a priori* thresholds for agreement will be set.

Ethical Approval

The proposed study has received full ethical clearance from a human and social sciences ethics committee.

Results

Initial results from the systematic review revealed a paucity of scientific literature that documented the objective assessment of hypotonia in children. The review identified the need for more studies with greater methodological rigor in order to determine best practice with respect to the methods used in the assessment of low muscle tone in the paediatric population.

Discussion

This proposed study will be guided by the overarching principles of evidence-based practice and will contribute to instrument development, more specifically with respect to a clinical algorithm. The study will also assist in addressing the current gaps in the literature, which may be considered the epistemological contribution. The study will additionally inform and provide a multidisciplinary team (paediatricians, occupational therapists, physiotherapists) with a clinical tool that is based on evidence and validated. It will also provide knowledge into the best practices for the assessment of low muscle tone to address current diagnostic challenges, and finally will contribute empirical evidence towards shaping educational curricula in the absence of a gold standard.

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Conflicts of Interest

This study is supported by the Medical Research Council of South Africa in terms of the National Health Scholars Programme from funds provided for this purpose by the National Department of Health.

Multimedia Appendix 1

MRC Scholarship/Grant Letter.

[PDF File (Adobe PDF File), 88KB - resprot_v3i4e71_app1.pdf]

Multimedia Appendix 2

NRF Grant Award Letter.

[PDF File (Adobe PDF File), 215KB - resprot_v3i4e71_app2.pdf]

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Abbreviations

AGREE-II: Appraisal of Guidelines for Research & Evaluation II
ATLAS-ti: Archiv fuer Technik, Lebenswelt und Alltagssprache-text interpretation
ICF: International Classification of Functioning
ISO: International Organization for Standardization

ITU-T: International Telecommunication Union
SPSS: Statistical Package for the Social Sciences

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Protocol

Development and Validation of a Personalized, Web-Based Decision Aid for Lung Cancer Screening Using Mixed Methods: A Study Protocol

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Abstract

Background: The National Lung Screening Trial demonstrated that low-dose computed tomography (LDCT) screening could be an effective way to reduce lung cancer mortality. Informed decision-making in the context of lung cancer screening requires that potential screening subjects accurately recognize their own lung cancer risk, as well as the harms and benefits associated with screening, while taking into account their personal values and preferences.

Objective: Our objective is to develop a Web-based decision aid in accordance with the qualifying and certification criteria in the International Patient Decision Aid Standards instrument version 4.0 that will assist patients in making informed decisions with regard to lung cancer screening.

Methods: In “alpha” testing, a prototype of the decision aid was tested for usability with 10 potential screening participants in focus groups. Feedback was also sought from public health and health risk communication experts external to the study. Following that, improvements to the prototype were made accordingly, and “beta” testing was done in the form of a quasi-experimental design—a before-after study—with a group of 60 participants. Outcomes tested were knowledge, risk perception of lung cancer and lung cancer screening, decisional conflict, and acceptability of the decision aid as determined by means of a self-administered electronic survey. Focus groups of a subsample of survey participants will be conducted to gain further insight into usability issues.

Results: Alpha testing is completed. Beta testing is currently being carried out. As of 2014 December 7, 60 participants had completed the before-after study. We expect to have results by 2015 January 31. Qualitative data collection and analysis are expected to be completed by 2015 May 31.

Conclusions: We hypothesize that this Web-based, interactive decision aid containing personalized, graphical, and contextual information on the benefits and harms of LDCT screening will increase knowledge, reduce decisional conflict, and improve concordance between patient preferences and the current US Preventive Services Task Force’s screening guidelines.

(JMIR Res Protoc 2014;3(4):e78) doi:10.2196/resprot.4039

KEYWORDS

informed decision-making; lung cancer screening; patient decision aid; patient education

Introduction

Lung cancer remains the leading cause of cancer death in the United States [1]. The National Lung Screening Trial demonstrated that lung cancer screening with low-dose computed tomography (LDCT) has the potential to significantly reduce lung cancer mortality [2]. On the basis of this and other evidence, the US Preventive Services Task Force (USPSTF) gave a B recommendation for LDCT screening [3,4], the same strength of recommendation associated with mammography screening. However, real-world success in lung cancer screening will be conditional on identifying and screening those at highest risk for lung cancer while discouraging screening in those at low risk. Lung cancer screening presents a challenge, because it is the first population-wide screening modality with eligibility criteria based not only on age but also on a lifestyle behavior (at least 30 pack-years of tobacco use and smoking within the past 15 years). Identifying those at risk and helping them understand the benefits of screening and how to reduce their risk (e.g., tobacco cessation) is paramount for an effective implementation of population-wide lung cancer screening.

Implementing lung cancer screening in an environment where patients do not have the tools or information to understand disease risks and the harm-benefit balance of screening will most likely be counterproductive. In addition to providing information for individuals regarding lung cancer screening that allows them to weigh the potential harms of LDCT in accordance with the benefits, we also recognize that the decision to be screened is preference-sensitive. In light of this, there is a need to assist individuals with making informed decisions regarding lung cancer screening in which their personal values are also taken into account.

The USPSTF defines informed decision-making as "an individual's overall process of gathering relevant health information from both his or her clinician and from other clinical and nonclinical sources, with or without independent clarification of values" ([5], p. 59). In particular, a patient decision aid's functions are (1) to provide facts about an individual's condition and the options available and their characteristics, (2) to help individuals clarify values and personal preferences, and (3) to assist these individuals to discuss their values and preferences with health professionals [6]. It is in this context that our goal is defined: to create a decision aid that improves the knowledge of LDCT screening, decreases decisional conflict, and improves concordance of screening preferences between the official recommendations and the individual (i.e., to assist individuals with informed decision-making about whether or not to screen). Concordance with official recommendations is important, because this will ensure that the resulting population of screened individuals is consistent with that for which lung cancer screening is deemed effective.

Evidence shows that decision aids can improve decision quality as a result of better knowledge of options and their associated harms and benefits; decrease decisional conflict; and reduce the overuse and increase the underuse of screening options [7-10]. In particular, Jimbo and colleagues [9] focused on cancer screening and outlined recommendations for the evaluation of decision aids, the most important of which was to base the decision aid on a theoretical framework so that relevant outcomes are measured. The development of the decision aid in our study uses the Ottawa Decision Support Framework, a widely used theoretical framework that applies theories from psychology, social psychology, economics, and social support [11].

Whereas numerous decision aids exist for prostate, colon, and breast cancers, there are only a handful of tools that fulfill the functions of a patient decision aid, either partially or fully, with regard to LDCT screening [12-16]. We are aware of only one that has formally evaluated measures of the effect of decision aids established in the theories of behavioral research in a peer-reviewed journal [16]. However, this decision aid [16] is in the format of a video that provides average risks and benefits of LDCT screening. We know that risk varies greatly among smokers [17]; therefore, in our study, we propose to develop an alternative format: a Web-based, interactive decision aid that takes into account a more accurate depiction of an individual's personal lung cancer risk. The decision aid will comply with the qualifying and certification criteria set out in the International Patient Decision Aid Standards instrument (IPDASi) version 4.0 [18]. The rationale for a Web-based tool is that the Web is increasingly becoming an important source of cancer information [19]. Another advantage of the Web-based format is that the content of decision aids could easily be expanded and customized for different audiences. Studies have also confirmed the feasibility of using the Web to deliver the Web-based decision aids [20].

Methods

Prototype Development

A prototype was developed based on the most recent clinical guidelines provided by the USPSTF [5], the IPDASi version 4.0 checklist [18], and recommendations in terms of risk communication [21-23]. The decision aid includes information on personalized lung cancer incidence risk calculated by using an established risk model [24], risk factors of lung cancer reported in the literature, harms and benefits of LDCT screening, and an explicit values clarification exercise. We also used a currently existent print-based decision aid developed by the Veterans Affairs Healthcare System as a reference [13,14].

The distinguishing factor of the Web-based version is that it allows individuals to compute their individualized lung cancer risk using an established risk model. Although all cancer screening is based on some risk factor identification, for the most common evidence-based cancer screening (e.g., colorectal, breast, and cervical cancer screening), risk factor identification involves little more than noting the relevant age and sex data. Lung cancer screening differs in very important ways in that proper application of current recommendations involves measuring risk by also identifying the individual's cumulative tobacco exposure measured in pack-years and the timing of tobacco use. Whereas smoking history accurately identifies risk on a population basis, it is less useful for individuals because there is such great variability of lung cancer risk, even among

smokers with similar exposure. Current models that more accurately quantify individual lung cancer risk incorporate up to 10 clinical and demographic variables that provide a more accurate, though complex, determination of risk. This requires sophisticated lung cancer risk prediction models [24].

In addition to accurately characterizing individual lung cancer risk, there is evidence that individuals may prefer to have such tailored information, which, in turn, may affect health-care-seeking decisions [10,25]. To this end, personalized risk in absolute terms (as a percentage) and individualized benefits in the form of icon arrays are generated by the decision aid. We also provide average harms as an estimation of individual harms (see Multimedia Appendix 1 for the current iteration of the results page).

We believe that putting LDCT screening in context with other common screening recommendations in terms of reduction of disease-specific mortality (i.e., reduction of breast cancer death by screening with mammography) will allow the individual to gauge how LDCT screening compares to other widely accepted screening practices. This has not been done with the available decision aids for LDCT lung cancer screening [13,14,16].

Participants

Our target population comprised of potential users of LDCT screening. The specific inclusion criteria are given in Textbox 1.

These criteria applied to all 3 phases of the study detailed below: Phase I alpha testing focus groups, Phase IIa beta testing before-after survey, and Phase IIb beta testing postsurvey focus groups.

Textbox 1. Inclusion criteria.

- Current and former smokers
- Aged 45-80 years
- No history of lung cancer
- No previous chest computed tomographic scan in the past year

Recruitment Procedure

A combination of passive and active recruitment was done to form 2 convenience samples of participants: 10 for alpha testing and 60 for beta testing. An advertisement was placed on the University of Michigan's (UM's) online portal for volunteers of clinical studies [26], along with flyers at all of the clinics in the University of Michigan Health System (UMHS) and district libraries in Ann Arbor. Active recruitment involved in-person recruitment at a pulmonary clinic and general internal medicine clinic in the UMHS. Also, a list of participants and their contact details was generated by the Honest Broker Office of UM Medical School from medical records who fulfilled the inclusion criteria as listed above. The project coordinator verified the eligibility of all potential participants over the phone.

Phase 1: Alpha Testing

Overview

We solicited feedback from public health and health risk communication experts with regard to the content and wording and how risk is expressed in the prototype. After incorporating their suggestions, we conducted focus groups with potential users of LDCT screening to test the usability of the prototype as part of the decision tool's iterative development process. Specifically, we pretested our tool for comprehension of the content, as well as the acceptability of the design, layout, and messages conveyed. A total of 10 people participated in 1 of 2 focus groups. The eligible and willing participants were sent a Web link to work through the Web-based decision aid prototype via e-mail. All participants were asked to participate in the focus group within the week they reviewed the decision aid. Version 1 was the product of alpha testing.

Data Collection

The focus group was conducted by a trained external facilitator and a study team member using an interview guide developed by study team members with expertise in qualitative research and lung cancer screening decision-making (see Multimedia Appendix 2). Focus groups were held at a venue at the UM School of Public Health. At the focus group, individuals were asked to complete a survey to document the demographic makeup of the focus group participants (see Multimedia Appendix 3). Thereafter, all participants accessed the decision aid again on an iPad individually to refresh their impression. After all participants in the focus group were done reviewing the tool, the external facilitator asked questions pertaining to the usability of the decision aid. All sessions were audiotaped, and field notes were taken. Ten minutes before the scheduled end of the focus group, the participants were asked to fill out an exit survey with 4 questions (see Multimedia Appendix 4).

Analysis

Audio recordings were transcribed verbatim. Data analysis took place simultaneously with data collection, which, in turn, assisted in the iterative development of both the interview guide and decision aid. Using the transcriptions and field notes, a brief report was given by the study team member present at the focus groups to the rest of the study team. The study team then decided what changes to incorporate into the decision aid, forming version 1.

Phase IIa: Beta Testing: Self-Administered Electronic Survey

Overview

Following alpha testing, we conducted a pilot study of version 1 of our decision aid with 60 individuals using a

quasi-experimental design: a before-after study. This study design is consistent with the development of a decision aid and is an accepted method to test the effectiveness of decision aid tools under "real-life" conditions [16,27,28]. The before-after study will be followed by focus groups to gain further insight into the outcomes of interest and other spontaneous feedback about the tool in general.

Data Collection

Following a successful screen for eligibility, a participant was invited to come to UM to complete a series of surveys administered on a computer by Qualtrics, an online survey tool. In particular, a participant began with the knowledge survey (see Multimedia Appendix 5). This was followed by the pretest demographic, lung cancer risk, and prior screening experiences survey (see Multimedia Appendix 6); risk perception of lung cancer and lung cancer screening (see Multimedia Appendix 7); and the decision conflict scale survey (see Multimedia Appendix 8). This formed the "before" survey, and the participant was automatically redirected to the website upon clicking "submit." The participant was instructed to explore the website for 10-15 minutes and on the final page, he or she would click on a link at the bottom that would redirect them to the "after" survey. The "after" survey comprised the knowledge, risk perception of lung cancer, and lung cancer screening; the values clarification questionnaire (see Multimedia Appendix 9); the decision conflict scale; and the acceptability survey (see Multimedia Appendix 10). The whole process lasted approximately 45 minutes.

Analysis

The outcomes measured were adapted from the Ottawa Decision Support Framework: knowledge of the benefits and risks of lung cancer screening, acceptability [29], decisional conflict [30], and concordance between the USPSTF's recommendation and an individual's preference. Concordance is a binary variable defined as "Yes" = 1, where an individual prefers to get screened (or not to get screened) and is eligible (or ineligible) and "No" = 0 if an individual prefers not to get screened (or to get screened) but is eligible (or ineligible). We also measured a participant's risk perception as recommended by relevant literature [9]. To compare before-after outcomes, we will use the Wilcoxon signed-rank test for continuous outcomes (knowledge and decisional conflict) and McNemar's test for binary outcomes (concordance and risk perception indicators). Frequency statistics will be computed for the acceptability items, as these will be measured only in the "after" survey. The software Stata 13 will be used to carry out all analyses [31].

Phase IIb: Beta Testing: Focus Group

Overview

Following completion of the survey, participants were asked if they would like to participate in a focus group to allow them to give the study team more feedback about the decision aid they just viewed. If the answer was affirmative, the participant was told that he or she could be contacted within the month to make an appointment. They were also asked whether the decision aid had indicated that they were eligible for screening (based on USPSTF guidelines) and what the risk of dying due to lung cancer was as computed by the calculator.

Focus groups will be stratified by self-reported eligibility for LDCT screening. Given that one of the main goals of the tool is to increase concordance with USPSTF eligibility guidelines, it will be useful to have specific focus groups consisting of screen-(in)eligible individuals entirely. The aim is to have 4-8 individuals per focus group. Where possible, focus groups will also be stratified by sex and age, given eligibility for LDCT screening.

Data Collection

The same steps will be followed for beta testing as those used for the focus groups conducted for alpha testing. The current version of the interview guide can be seen in Multimedia Appendix 11.

Analysis

Thematic analysis will be done on the data yielded from the focus group sessions. Two study team members with qualitative analysis expertise will code the data separately and compare codes to establish the themes to be explored. A report will be given to the rest of the team, and all members will decide what changes to include for the next iteration of the decision aid.

Research Ethics

All study participants will be asked to complete consent forms. Specific consent for audiotaping of focus groups will also be sought. This study was approved by the University of Michigan Medical School Institutional Review Board on 2014 June 18 (Study ID: HUM00088232).

Results

Alpha testing is completed. Beta testing is currently being carried out. As of 2014 December 7, 60 participants had completed the before-after study. We expect to have results by 2015 January 31. Qualitative data collection and analysis are expected to be completed by 2015 May 31. The current iteration of the results page with personalized risk generated by the decision aid can be seen at Multimedia Appendix 1.

Discussion

Preliminary results from the before-after study indicate that the decision aid improves knowledge about lung cancer screening, decreases decisional conflict, and increases concordance between USPSTF recommendations and the screening option preferred by the user. Therefore, we anticipate that the decision aid will be helpful to individuals in making informed decisions about lung cancer screening.

Acknowledgments

YKL, TJC, DA, and RM conceived of the study and sought funding and ethical approval. YKL and RM are responsible for the management of the alpha and beta testing phases and planned the statistical analysis. STC and PC assisted in the development of the prototype and subsequent iterations of the decision tool. PC and MW have assisted with data collection in both phases of testing. All authors have been involved in drafting and revising the manuscript and approved the final version.

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Conflicts of Interest

None declared.

Multimedia Appendix 1

An example of the current iteration of the results page.

[PNG File, 256KB - resprot_v3i4e78_app1.png]

Multimedia Appendix 2

Focus group interview guide - alpha testing.

[PDF File (Adobe PDF File), 53KB - resprot_v3i4e78_app2.pdf]

Multimedia Appendix 3

Focus group participant survey.

[PDF File (Adobe PDF File), 60KB - resprot_v3i4e78_app3.pdf]

Multimedia Appendix 4

Focus group exit survey.

[PDF File (Adobe PDF File), 17KB - resprot_v3i4e78_app4.pdf]

Multimedia Appendix 5

Before-after survey: knowledge.

[PDF File (Adobe PDF File), 70KB - resprot_v3i4e78_app5.pdf]

Multimedia Appendix 6

Before-after survey: demographics and risk factors.

[PDF File (Adobe PDF File), 59KB - resprot_v3i4e78_app6.pdf]

Multimedia Appendix 7

Before-after survey: risk perception.

[PDF File (Adobe PDF File), 33KB - resprot_v3i4e78_app7.pdf]

Multimedia Appendix 8

Before-after survey: decisional conflict.

[PDF File (Adobe PDF File), 50KB - resprot_v3i4e78_app8.pdf]

Multimedia Appendix 9

Before-after survey: values clarification.

[PDF File (Adobe PDF File), 49KB - resprot_v3i4e78_app9.pdf]

Multimedia Appendix 10

Before-after survey: acceptability.

[PDF File (Adobe PDF File), 33KB - resprot_v3i4e78_app10.pdf]

Multimedia Appendix 11

Focus group interview guide – revised for beta testing.

[PDF File (Adobe PDF File), 63KB - resprot_v3i4e78_app11.pdf]

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Abbreviations

IPDASi: International Patient Decision Aid Standards instrument
LDCT: low-dose computed tomography
NLST: National Lung Screening Trial
UM: University of Michigan
UMHS: University of Michigan Health System
USPSTF: US Preventive Services Task Force

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Original Paper

Developing Community-Based Rehabilitation Programs for Musculoskeletal Diseases in Low-Income Areas of Mexico: The Community-Based Rehabilitation for Low-Income Communities Living With Rheumatic Diseases (CONCORD) Protocol

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Abstract

Background: The negative impact of musculoskeletal diseases on the physical function and quality of life of people living in developing countries is considerable. This disabling effect is even more marked in low-socioeconomic communities within developing countries. In Mexico, there is a need to create community-based rehabilitation programs for people living with musculoskeletal diseases in low-socioeconomic areas. These programs should be directed to prevent and decrease disability, accommodating the specific local culture of communities.

Objective: The objective of this paper is to describe a research protocol designed to develop, implement, and evaluate culturally sensitive community-based rehabilitation programs aiming to decrease disability of people living with musculoskeletal diseases in two low-income Mexican communities.

Methods: A community-based participatory research approach is proposed, including multi and transdisciplinary efforts among the community, medical anthropology, and the health sciences. The project is structured in 4 main stages: (1) situation analysis,

(2) program development, (3) program implementation, and (4) program evaluation. Each stage includes the use of quantitative and qualitative methods (mixed method program).

Results: So far, we obtained resources from a Mexican federal agency and completed stage one of the project at Chankom, Yucatán. We are currently receiving funding from an international agency to complete stage two at this same location. We expect that the project at Chankom will be concluded by December of 2017. On the other hand, we just started the execution of stage one at Nuevo León with funding from a Mexican federal agency. We expect to conclude the project at this site by September of 2018.

Conclusions: Using a community-based participatory research approach and a mixed method program could result in the creation of culturally sensitive community-based rehabilitation programs that promote community development and decrease the disabling effects of musculoskeletal diseases within two low-income Mexican communities.

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KEYWORDS

rehabilitation; community-based participatory research; social change; musculoskeletal diseases; Mexico

Introduction

Musculoskeletal Diseases and Disability

Musculoskeletal diseases are highly prevalent in communities of many developed and developing countries, resulting in important health problems for individuals and society [1-4]. Many epidemiological studies performed in developed countries have found high levels of disability and work absenteeism among people who suffer musculoskeletal disorders [5-8]. Furthermore, disability produced by musculoskeletal pain has a negative impact on the social and emotional well-being of people [9], especially in the older adult population [10].

The negative impact of musculoskeletal diseases on the physical function and quality of life of people is more marked within developing countries [11]. This could be linked to observations that increased disability is associated with lower socioeconomic levels [12,13]. A large epidemiological study conducted in Mexico reported that the prevalence of musculoskeletal pain was 26%, which was associated with 13% of physical disability [14]. This study also found significant differences in the regional prevalence of musculoskeletal pain and its causes across the country, implying the influence of different cultural, socioeconomic, and demographic factors within each geographical location [14].

In the Mexican northern state of Nuevo León, the prevalence of osteoarthritis is 17% [15], while the estimated national prevalence is 10.5% [14]. This shows that osteoarthritis is an important health problem for this region. In the southern state of Yucatán, the existence of chronic musculoskeletal diseases, such as osteoarthritis, back pain, and rheumatoid arthritis, produces a 6% prevalence of disability negatively affecting the life of the people living in this region [16,17]. As a result, there is a growing interest in designing community-level interventions directed to decrease the musculoskeletal-related disability within these Mexican regions.

Rehabilitation Interventions

Specifically, the health professionals of the University Health Center of Nuevo León (UHC-Nuevo León) have a particular interest in addressing the health problems posed by musculoskeletal diseases in their community. The UHC-Nuevo León is a primary health care program run by the Autonomous University of Nuevo León that provides health services to a large community of low socioeconomic level. On the other hand, the Latin American Group for the Study of Rheumatic Conditions in Indigenous People (Grupo Latinoamericano para el Estudio de Enfermedades Reumaticas en Poblaciones de Origen, GLADERPO) is interested in creating interventions for decreasing the disabling effects of musculoskeletal diseases in a municipality called Chankom, which is an underserved Mayan community located in the state of Yucatan. Consequently, these two groups are looking to design rehabilitation interventions aimed to address the musculoskeletal-related disability within their communities of interest.

Rehabilitation is defined as an “enabling” process aimed at reversing the “disabling” effects of a pathological condition [18] or a social situation [19]. This process involves efforts directed both at the persons and their environments, allowing them to get “back on track” with their lives and to achieve equal opportunities to participate in their desired social roles [20]. There is evidence that rehabilitation is effective at reducing the burden of disability, enhancing opportunities for disabled people. This results in an improvement of quality of life to the extent that the United Nations and the European Board of Physical and Rehabilitation Medicine consider “access to rehabilitation” as a human right [21,22].

Particularly, rehabilitation interventions have proven effective to decrease pain and improve physical function with people suffering from rheumatologic diseases [23]. Nevertheless, in Mexico only 1.7% of the people who suffer from musculoskeletal diseases receive rehabilitation [14]. Consequently, there is a need to develop community rehabilitation programs directed at decreasing the disabling effects of musculoskeletal diseases in both the community served by the UHC-Nuevo León (community-UHC-Nuevo León) and the Mayan community of Chankom.

Community-Based Rehabilitation and Community-Based Participatory Research

The concept of community-based rehabilitation (CBR) has evolved over 30 years of community work, mostly in developing countries. CBR started as an approach of biomedical service and gradually progressed to a “human-rights” approach

supporting community development [24]. Therefore, this approach is now defined as a community development strategy for the social inclusion of people with disabilities through the equalization of opportunities [25]. Due to its participatory focus it has been proposed that CBR is a "democratic tool for social change" [26].

Nevertheless, there have been some limitations in the application of the CBR approach worldwide, which include a lack of cultural sensitivity [24]. Cultural sensitivity refers to the ability to accommodate a specific culture [27], and successful community programs address this by including the knowledge, beliefs, and values of the target community [28]. Therefore, CBR programs should be culturally sensitive; in other words they need to be developed with primary consideration of the beliefs, perceptions, and values of the culture of the community where they will be implemented.

The concept of cultural sensitivity obtains significant relevance when dealing with very different communities, as in the case of the community-UHC-Nuevo León and Chankom. The 5 community health centers that form the community-UHC-Nuevo León provide care to 52 urban neighborhoods (approximately 140,000 persons). The entire population of this community speaks Spanish and belongs to a low to middle-low socioeconomic level. On the other hand, the community of Chankom Municipality has 4340 inhabitants spread across 11 small rural settlements or commissariats. The majority of Chankom's population speaks Mayan and lives in very high levels of poverty. Given the sociocultural differences between these two communities, it is essential to adopt the concept of cultural sensitivity, and not to take a "one size fits all" approach for the development of the CBR programs.

Another important limitation of the CBR approach is the lack of formal research and scientific evaluation of its goals and processes [24,29-31]. Culturally sensitive CBR programs can be achieved through a "full and effective participation of the community" [25]. Consequently, participatory research strategies could represent a viable alternative to do research on CBR. There is one strategy, the community-based participatory research (CBPR), which has been proposed as an optimal method to develop culturally sensitive community-based health programs [32]. This strategy is part of the "participatory action" research that conceptualizes reality as formed by objective and subjective perspectives. Perspectives are historically constituted and reconstituted by human agency and social action, which implies a need to establish a dialectic relationship among different forms of knowledge production [33]. As a result, the CBPR approach involves the use of different quantitative and qualitative strategies to generate knowledge, which then can be used to address community needs [34,35].

CBPR is based on the following principles: (1) acknowledgment of the community as a unit of identity, (2) development of community strengths and resources, (3) promotion and facilitation of equitable and participatory partnerships with community members in all phases of research, (4) promotion of colearning and capacity building for all partnership members, (5) achievement of balance between knowledge generation and intervention for the mutual benefit of all partners, (6) focus on relevant problems for the community, (7) use of iterative and cyclical processes in all research, (8) involvement of all partners in the local and global dissemination of results, and (9) establishment of long-term commitment with partnership sustainability [32]. The application of these principles can result in knowledge that is owned by the community and is useful for the design, implementation, and evaluation of community interventions [32,35].

The use of CBPR strategies has resulted in increments of community capacity and positive effects on community health [36]. In Latin America, there is a long history of health-program development efforts through social participation, which have repeatedly failed to achieve all their goals [37]. Lessons learned from these experiences suggest that collaborative efforts established between communities and nongovernment institutions, such as universities, are an efficient way to solve immediate health issues, improve resource utilization, and raise social and political awareness [37]. In consequence, a CBPR strategy that includes alliances between community and academic institutions could be effective, producing structured social participation to solve disability related problems in the community-UHC-Nuevo León and the Municipality of Chankom.

Main Intention and Objective

It has been stated that what really defines a social participation approach are the intentions and meanings given to the actions conducted by the people involved in it, and it is extremely important to be transparent about the intentions of using such a research strategy [38]. The main intention of this project is to organize and empower communities to develop a culturally sensitive CBR (csCBR) program in partnership with academics. This partnership will seek to form alliances with government and nongovernment institutions in order to ensure the success and continuation of the program. The csCBR program will aim to reduce the disabling effects of musculoskeletal diseases through supporting the efficient use of resources available in the communities and promoting micro and macro social changes.

The main objective of this protocol, which we named Community Based Rehabilitation for Low Income Communities Living With Rheumatic Diseases (CONCORD), is to develop, implement, and evaluate csCBR programs to decrease disability of people living with musculoskeletal diseases in the community-UHC-Nuevo León and the Municipality of Chankom. The hypothesis of this project is, "The execution of a CBPR strategy that permits a fusion of global and local knowledge will result in the creation of csCBR programs that will promote community development, thereby increasing social integration of disabled people with musculoskeletal diseases living in the communities of interest".

Theoretical Approach

The theoretical approach of this research project aligns with a social constructivist worldview, assuming that a successful CBR program can be developed through the construction of "new knowledge". This new knowledge results from the "fusion of horizons" [39] between global knowledge (scientific/academic) and local knowledge (community beliefs and values). The new

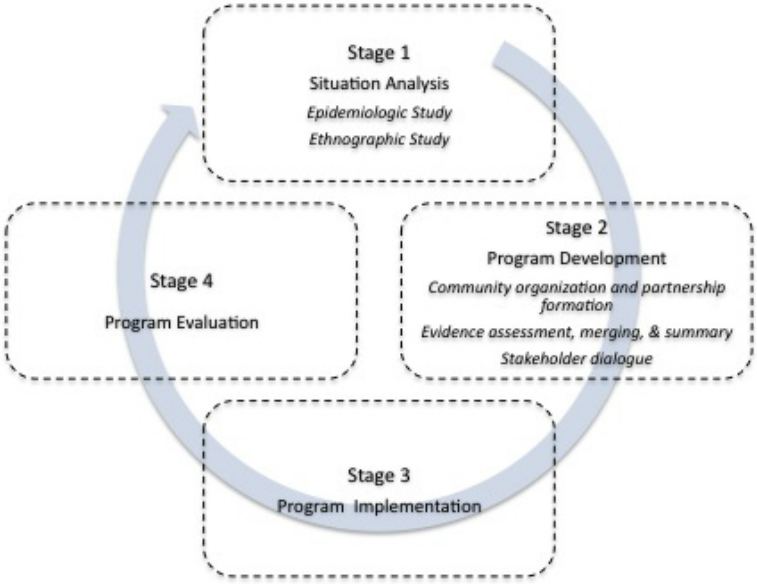
knowledge will be supported by community and academic members and will permit the definition of actions to facilitate its use for the benefit of the community. These actions will be structured as a complex intervention [40] in the form of a csCBR program and will involve collaborations with representatives of social and health policy institutions. In addition, we will use critical analytic approaches to disclose and resolve conflicts of power innate to every participatory action project.

Methods

Research Strategy and Methodology Overview

The CBPR strategy in this project will include a multi and transdisciplinary effort that involves a dialogic relationship between medical anthropology and some health sciences such as rheumatology, epidemiology, rehabilitation, nursing, and primary health care. Following the 2010 World Health Organization (WHO) guidelines for the development of CBR programs [41], this project is structured in four main stages (Figure 1 shows these stages). Methodologically, this project is conceived as a mixed method program, which involves the use of quantitative and qualitative methods in all its stages [42]. Following is a description of the main methodological elements that constitutes each of these stages. Differences on how each stage will be conducted in each of the communities of interest will also be noted.

Figure 1. The "Community-Based Rehabilitation for Low Income Communities Living With Rheumatic Diseases (CONCORD)" protocol.



Stage One: Situation Analysis

Objectives

This stage will be undertaken over a 6-month period with the objectives of: (1) generating knowledge about the physical function problems produced by musculoskeletal diseases in the target communities; and (2) understanding the specific contexts in which these problems occur within each community. To achieve these objectives we will conduct an epidemiologic study in parallel with an ethnographic study.

Epidemiologic Study

This will be a "pure quantitative" study [42] with the specific objectives of: (1) assessing the prevalence and factors associated with musculoskeletal diseases development and progression in both communities of interest; and (2) evaluating the impact of musculoskeletal diseases on the physical function and health status of the people living in both communities. We expect to understand the impact of musculoskeletal diseases within the communities and to identify the presence of potentially modifiable factors to prevent or decrease disability.

This will be an observational, cross-sectional, survey-based study. Due to the different population sizes, in Chankom we will conduct a census of all adults (≥18 years old) living in the community; whereas in the community-UHC-Nuevo León we will obtain a multistage probabilistic sample of 1516 adults (considering a precision of 3%, a 95% confidence level, an estimated osteoarthritis prevalence of 20%, observed in Nuevo León State, and the sample size adjustment recommended for multistage sampling procedures) [43]. The primary sampling unit of the probabilistic sampling will be neighborhoods served by the UHC-Nuevo León. The secondary sampling unit will be blocks within selected neighborhoods, and the tertiary sampling unit will be households within selected blocks. All procedures will be performed using a random-start systematic proportional sampling procedure. In order to control for within-household homogeneity, we will only survey one person per household.

The survey procedure will be structured following the Community Oriented Program for the Control of Rheumatic Diseases (COPCORD) methodology [44]. The COPCORD is a screening strategy to detect rheumatologic disorders in the community and has proven to be effective when used in Mexico [45]. Briefly, the survey consists of a questionnaire designed

to explore the presence of joint pain, stiffness, and inflammation along with factors associated with musculoskeletal diseases, physical activity, physical function, and health status.

Physical activity will be assessed using the well validated Mexican-Spanish version of the Rapid Assessment of Physical Activity questionnaire [46,47]. Health status will be evaluated through directly asking the participant, "How have you been with your illness?", and physical function will be measured through the Health Assessment Questionnaire Disability Index (HAQ-DI). This questionnaire, which has shown good psychometric properties when applied to people with musculoskeletal chronic diseases [48], is available in Spanish [49] and has been validated within the Mexican population [50,51]. The survey will also include a socioeconomic assessment including education, income, home characteristics, and commodities.

Trained personnel will administer the survey to both communities in person. In the case of Chankom, a cross-cultural adaptation of the instrument to the Mayan language was conducted [52]. A trained general physician will assess all adults that reported any musculoskeletal symptoms at their homes using standardized criteria for the diagnosis of rheumatologic diseases. A specialist (rheumatologist or physiatrist) will confirm all cases identified with rheumatologic diseases.

The specialist will conduct a thorough medical assessment of all confirmed cases. This assessment will include radiographic evaluation, medical history, and physical examination with the objectives of evaluating the impact of disease on physical function and the presence of factors for functional decline and disease progression.

Physical function will be evaluated according to Glass's tenses of "human functioning" [53]. These tenses are: (1) "enacted tense" or performance of meaningful activities within life context; (2) "hypothetical tense" or perceived capacity to do predefined activities; and (3) "experimental tense" or capability to do activities in standardized conditions. Performance of meaningful activities will be evaluated by self-report of main housework, work, and leisure activities, including an assessment of the concept "preclinical disability". Preclinical disability refers to the state in which, in spite of no interruptions in the execution of regular activities, there is a modification of the way and/or the frequency in which these activities are performed [54]. Perceived capacity to do predefined activities will be assessed using the HAQ-DI, described above. Finally, capability to do activities in standardized conditions will be evaluated through the 6-minute walk test (6MWT) and the functional dexterity test (FDT). The 6MWT measures the distance an individual can walk during 6 minutes on a hard, flat surface [55], and has shown good test-retest reliability when used with people with musculoskeletal conditions such as osteoarthritis [56,57]. The FDT evaluates the ability to use the hand for "functional daily tasks that require 3-jaw chuck prehension between the fingers and the thumb" [58] and has shown good intra and interrater reliabilities and construct-validity in diverse pathologic conditions of the hand [59].

A member of the research team will perform periodic screenings to ensure the quality of the database. We will estimate descriptive statistics (central and dispersion estimates). In the case of the community-UHC-Nuevo León, we will also estimate 95% confidence intervals correcting for the three-stage sampling. We will use linear and logistic regression models to evaluate the factors associated with disease presentation and with impact on health and physical function utilizing specialized statistical software (STATA version 12).

Ethnographic Study

This "pure qualitative" study [42] will be conducted over 6 months, in parallel with the epidemiologic study, with the objective to produce an ethnographically informed report on the "explanatory models of illness" within the medical-anthropological "health systems" [60] of Chankom and the community-UHC-Nuevo León. Explanatory models of illness refer to the different narratives present on the causes, manifestations, trajectories, and treatments of disease, whereas the medical-anthropological health systems include the popular, traditional, and professional contexts in which health is conceived [61].

We will conduct a study from the perspective of ethnography [62]. This implies the conducting of fieldwork where anthropologists and other researchers-in-training will live in or close by the target communities. Given the high rates of violence registered in Nuevo León during the last few years, we will conduct preliminary in-depth interviews and focus groups with community leaders and health providers of the UHC-Nuevo León to ensure that it is safe for a researcher to live in this area, and in case it is not, to define alternative strategies to complete the planned fieldwork.

The fieldwork will include purposeful sampling of key persons, activities, social and familiar events, and documents. Key persons will include: (1) community members who have musculoskeletal diseases involving different body regions; (2) community leaders involved in community development activities; (3) representatives of health professionals involved in the care of people with musculoskeletal diseases in these communities; (4) representatives of health providers not officially recognized by a professional association (eg, bonesetters, masseurs, etc); (5) representatives of the local government; and (6) representatives of social development institutions (state and nongovernmental). These persons will be interviewed through informal and formal (in-depth interviews and focus groups) techniques.

We will perform participant and nonparticipant observations of individual's activities (eg, occupation) and social, familial, cultural, and provincial events. These observations will be chosen according to their relevance to the musculoskeletal disability problematic within each community. Finally, we will obtain written documents that are relevant to understand the problem of musculoskeletal disability within each community (eg, local disability laws, social welfare rules, clinical practice guidelines, advertisements, etc).

All activities in Chankom will be conducted using Mayan translators who are fluent in Spanish and Mayan languages and are recognized by the community as members of their own. Access to each community will be negotiated with community

leaders and local authorities. Data will be recorded by the use of field notes and audiotape recorders. We will aim to achieve thematic and/or theoretical saturation [63]. All data will be transformed to written electronic format and will be organized and managed using specialized qualitative data software (Hyperresearch, version 3.5.2).

Data will be analyzed and interpreted by the research team. The team will work on concept generation, typology development, and execution of comparative strategies. Constant reflection about team members' emotions and prejudices that emerge while conducting the fieldwork will be executed. Data analysis and interpretation will be done through a continuous cycle of analysis-interpretation-reflection. The analysis-interpretation phase will feed into the data acquisition phase; hence they will occur simultaneously. An iterative analytic-interpretative process will be use in which theoretical ideas will be used to make sense of data and the data will be used to change theoretical ideas [64]. All analytic, interpretative, and methodological decisions will be carefully registered as memoranda within an audit trail book.

Completing this ethnography will help us understand the disability problematic caused by musculoskeletal disorders in the communities of interest. This study will allow the identification of barriers and facilitators for the optimal function of the population who suffers from musculoskeletal diseases in Chankom and the community-UHC-Nuevo León. Understanding the local culture and the native perspective on the causes, management, impact, and prognosis of musculoskeletal diseases will help us define better the problematic related to musculoskeletal diseases within the communities. In addition, knowing the communities' local, regional, and national social structures along with their functional dynamics will orient us on how to proceed during the following stages of the project.

Stage Two: Program Development

Objectives

This stage will take 12 months to complete and has the following objectives: (1) to organize the communities and form a partnership among these and members of academia under the principles of equity and mutual respect; (2) to define the priority problems related to the disabling effects of musculoskeletal diseases, and to identify possible solutions to these problems; and (3) to define the components of the csCBR program along with the necessary actions to implement them, assuring the necessary resources to execute them. This stage will follow a "qualitative dominant" methods perspective [42] being composed of three main and sequential activities: (1) community organization and partnership formation; (2) evidence assessment, merging, and summary; and (3) stakeholder deliberation. In addition, an anthropologist will conduct ethnographic work including nonparticipant observations and in-depth interviews on all these activities in order to produce a reflective-critical analysis from a medical anthropology perspective.

Community Organization and Partnership Formation

We will present the information gathered during the initial stage of this project to the community through the organization of community meetings at different strategic locations. During these meetings we will form 2 types of committees labeled as "first-level" or "second-level" committees. In Chankom, we will hold 11 meetings, one at each commissariat, and in the community-UHC-Nuevo León we will conduct 5 meetings, one at each of the health care units that form this center. These information meetings have the goal of creating awareness about the disabling effects of the musculoskeletal conditions explored within these communities. By the end of each meeting we will ask the community to choose 4 persons to constitute a first-level committee. A person from each of these first-level committees will participate in the second-level committee. There will be only one second-level committee, which includes representatives of all the strategic locations within our target communities (11 in Chankom and 5 in the community-UHC-Nuevo León).

The second-level committee of each community will be legally constituted as a "civil association". This will be important for allocating and requesting financial resources, because in Mexico most government and nongovernment institutions can only serve organizations of this kind. The second-level committee will directly interact and work with representatives of the academic institutions involved in this project. During the first meeting of all committees, the members will define their roles as well as the rules for collaboration in relation to the processes of communication, decision making, and conflict resolution. We will use a nominal group technique, which is a group decision-making method, based on procedures for ideas' exposition, discussion, and ranking that allows everyone's opinion to be taken into account, reaching the best possible solution that is constituted by a mixture of all group members' ideas [65].

The second-level committee and the academics will be in charge of all methodological and administrative decisions for the project, as they will take on the role of the principal investigator. All decisions taken within this partnership between communities and academia, from now on referred to as "the partnership", will be the result of an ongoing analytic-interpretive-consensus process. In addition, the information and decisions generated within the partnership will be disseminated to the community via the first-level committees. In the same token, the community will be able to communicate with the second-level committee and academics through the first-level committees.

Evidence Assessment, Merging, and Summary

The first task for the partnership and the first-level committees will be to define the priority problems within their communities. The groups will use the knowledge generated during stage one of this project and the elements described by the WHO CBR matrix [41]. Priority problems refer to those issues that need to be urgently solved in order to decrease the disabling effects of musculoskeletal diseases at Chankom, and the community-UHC-Nuevo León. These issues will be organized and structured according to their main content in: (1) health, (2) education, (3) livelihood, (4) social, and (5) empowerment problems. The prioritization of problems will be based on their impact on the community's health and physical function. We will then think about possible solutions using both, the communities' social and cultural knowledge (local evidence) and the knowledge generated within the "scientific-academic"

world (global evidence). These ideas will redefine community priority problems based on the cost, benefit, and efforts required to implement them.

The global evidence assessment will largely be the responsibility of the academic partners. This will be accomplished by combining the methodology for "overview of reviews" proposed by the Cochrane Collaboration [66] and the "evidence assessment" approach proposed by the "Grading of Recommendations Assessment, Development and Evaluation" group [67]. Once processed, this evidence will be formatted into a fourth grade level of comprehension, so every member of the partnership and committees can understand it. Following this, the partnership will merge both local and global evidences in order to construct a plan to solve the disabling problems posed by musculoskeletal diseases in the communities. Therefore, it is expected that this plan will be both solid, in relation to its scientific foundation, and sensitive to the cultural and social realities of each of the target communities.

Priority problems and the plan to attend them will be defined and written as an evidence brief (ie, a document that summarizes how the available evidence pertains to a pressing problem, select options for addressing the problem, and key implementation considerations). This evidence brief will be structured following the ideas developed by the McMaster Health Forum [68,69], along with ideas from the "scenario planning" strategy for organization planning [70]. These briefs will include: (1) a clear description of each problem including its context; (2) a description of possible individual, community, programmatic, and systemic solutions to address each problem through the use of different scenarios; (3) a description of expected outcomes (benefits, costs, and harms) for each scenario; (4) a simple description of the grade of uncertainty behind the expected outcomes of each scenario; (5) a description of possible barriers for the implementation of each possible solution; and (6) a clear description about the sources from which the information of the possible solutions and scenarios came.

Stakeholder Dialogue

The components of the csCBR program will be defined using the principles of the Communicative Action Theory, which assumes that communication aimed at reaching agreement is the base from which to coordinate the activities of social change [71]. Consequently, we will create a space for communication or forum to convene a stakeholder dialogue to support action for improving health outcomes through collective problem solving by different key decision makers. The stakeholder dialogue will be conducted based on the methods developed by the McMaster Health Forum [72]. In addition, we will attempt to achieve an "unforced consensus" [33], a goal not usually targeted by these kinds of forums. This consensus will be fundamental to assure the execution and sustainability of the csCBR program.

Key decision makers are defined as those knowledge users who are able to influence the decision-making processes of their respective areas. The partnership will identify key decision makers using the information gathered during the previous stage and substages of the project. We anticipate that identified key decision makers will represent at least one of the following areas: (1) traditional medicine, (2) professional health care, (3) government and nongovernment social welfare, and (4) health policy. During this part of the project we will intend to form an alliance with these key decision makers in order to create commitments that will ensure human and material resources for the execution of the csCBR program, independently from resources of this research project. We will recognize these key decision makers as "powerful allies", based on the privileged position of power they held within their respective areas. Potential powerful allies will be invited to participate in the stakeholder dialogue through letters and person-to-person invitations.

The dialogue will be conducted over the course of several sessions in which participants will gather in a neutral, public location to talk about the information described in the evidence brief. A neutral facilitator, who will ensure a respectful and equitable communication among participants, will moderate the stakeholder dialogue. This facilitator will be responsible for all participants having the same chance to express their views during the dialogue. The final products from the stakeholder dialogue will include a dialogue summary (ie, a distillation of the key themes and insights that emerged during the dialogue) and the formation of a complex csCBR program composed by different components or actions along with a clear description of their respective expected outcomes. It is anticipated that these actions will include individual, community, and societal targets.

The components of the csCBR program will be defined through a nonforced consensus achieved through a process agreed on by all participants at the beginning of the dialogue. Once the dialogue is completed, the csCBR program will be written, and the resulting document will be shared with all participants in order to assure its fidelity in relation to what was agreed during the dialogue. Agreements with powerful allies will be confirmed and clinched by signing letters of commitment. This strategy aims to favor the long-term sustainability of the csCBR program within each targeted community.

Stage Three: Program Implementation

This stage will be completed over 6 months following a "quantitative dominant" approach [42]. The stage involves conducting a pilot test of the CBR program developed during stage two, and the implementation of an improved CBR program in the two communities of interest. The pilot test will help in identifying barriers and facilitators for the program's implementation, allowing corresponding program adjustments. All partnership members will contribute to the design, execution, and interpretation of the results of this stage.

We will choose 1 strategic site at each community (ie, 1 commissariat in Chankom and 1 health center of the UHC-Nuevo León) to implement the csCBR program designed during stage two of this protocol. An anthropologist will assess the operational aspects of the csCBR program using nonparticipant observations, informal interviews, in-depth interviews, and focus groups. This qualitative information will be used to design two questionnaires to evaluate the presence of facilitators and barriers for the implementation of each of the components of the csCBR program in the community. There will be one questionnaire designed for users of the program and

another one for personnel involved in the program's execution. Trained interviewers will apply the questionnaires to all participants of the pilot test through home visits, visits at jobsites, or telephone calls.

Qualitative data will be analyzed and interpreted by the anthropologist and some members of the partnership using content and thematic analysis techniques. This analysis then will be presented to all partners to decide the content of the questionnaires. We will use descriptive statistics to rank the frequency of facilitators and barriers observed during the pilot test. The partnership will use this information to make decisions about relevant changes to the original csCBR program and to elucidate implementation strategies aiming to improve its successful implementation in the community. Once changes have been made, we will proceed to implement the updated csCBR program in both communities.

Stage Four: Program Evaluation

This stage will last for 18 months following a "pure mixed methods" approach [42] implying the simultaneous execution of quantitative and qualitative methods, each one producing results that will converge in a complete explanation of the researched phenomenon [73]. The objectives of this stage are: (1) to understand which components of the csCBR program are more effective, and what are their mechanisms of action; and (2) to evaluate the impact of the csCBR program on the functioning and quality of life (QoL) of the people living with musculoskeletal diseases in Chankom and the community-UHC-Nuevo León. This stage will allow us to get a complete explanation and understanding about the impact and mechanisms of action of the csCBR program developed.

Quantitative methods will consist of a longitudinal, prospective, and comparative pre/post intervention observational design. Qualitative data will be gathered through ethnographic fieldwork to understand the dynamics and mechanisms of action of each of the csCBR program components. The ethnographic work will also inform quantitative findings about the impact of the program on functioning and QoL.

The quantitative sampling strategies will vary between our two target communities. In Chankom, we will include all the people enrolled in the CBR program together with a sample of people with equivalent ethnic, cultural, and socioeconomic characteristics, who live outside Chankom and have not been exposed to the program (control population). In the community-UHC-Nuevo León, we will assemble a random probabilistic sample of people with musculoskeletal diseases who are involved in the CBR program, and an equal sample of people with osteoarthritis living in a community with similar socioeconomic and cultural characteristics as the community-UHC-Nuevo León, but that has not been in contact with the program (control population). Quantitative results of stage one will provide us with the information needed to calculate appropriate sample sizes. The ethnographic work will require purposeful sampling of people who participated in activities that were implemented in the CBR program for at least 3 months, in both target communities. This will assure that sufficient experience with the program's processes and activities has been accumulated.

For the quantitative part, we will take baseline measurements, prior to the implementation of the program, and follow-up measurements every 6 months (4 measurements in total until 18 months) in both the target and control populations. Subjects of the control populations will be identified using the COPCORD screening methodology described in stage one of the project. We will measure: (1) 3 different tenses of physical function [53]; (2) QoL; and (3) outcomes related to each component of the csCBR program, whatever these may be.

As already mentioned, we anticipate that the csCBR program will include interventions at different levels, from the personal to the institutional level. In consequence, outcomes will be defined and measured according to the theoretical understanding of each level.

Hypothetical functioning will be measured through the WHO Disability Assessment Schedule 2 (WHODAS 2.0). The WHODAS 2.0 is a generic health-related disability assessment with excellent psychometric properties and was created through an extensive multicultural effort [74]. Experimental functioning will be evaluated using the 6MWT and the FDT. Both tests have shown excellent psychometric properties in musculoskeletal disease populations [57,59]. Enacted functioning will be measured subjectively through the Patient-Specific Functional Scale, which has shown excellent validity and reliability properties when applied in musculoskeletal-related pain populations [75], and semiobjectively using self-report, nonparticipant observations, and videos. QoL will be assessed through the WHO QoL Instrument. This instrument was developed through a multicultural collaboration and has been used with different populations, including older adults, showing excellent reliability and validity properties [76]. All questionnaires will be translated and culturally adapted to the Mayan language.

The ethnographic fieldwork will be conducted by a medical anthropologist and will include participant and nonparticipant observations, in-depth interviews, and focus groups. These qualitative methodologies will be conducted to understand the mechanisms of action of the different components of the program, along with their respective positive and negative aspects. In addition, the fieldwork data will help us in identifying relevant effects of the csCBR program, which can be measured quantitatively.

We will include descriptive and inferential statistic techniques to analyze the quantitative data. Inferential techniques will include multilevel modeling to explore effect modifiers on the outcomes of interest at different levels (eg, municipality, commissariat, or household levels), including between-group comparisons among target and control populations. We will use the statistical software STATA version 12. Ethnographic data will be analyzed following an analytic-interpretative-reflexive strategy from a medical anthropology perspective. These analyses will be further enriched by discussions with the partnership. All analytic and methodological decisions will be carefully registered in an audit trail. The results of this stage four will support decision-making processes within the partnership, allowing planning and conducting of a new situational analysis, thus completing the cyclical nature of the

project (see Figure 1). The cyclical nature of this project implies that the csCBR program's components will be constantly refined, and the outcomes expected by their implementation will be obtained after the execution of several cycles.

Results

The complexity of this project poses challenges for obtaining funding. Funding agencies in the developing world lack awareness of the need for this type of project and knowledge about the use of mixed methodologies. As such, we used different strategies for communicating the methods of the project to different audiences. In addition, we have applied for funding at diverse agencies, asking separate support for conducting the different parts of the project.

So far, we obtained resources from a GLADERPO study, founded by a Mexican federal agency, and completed stage one of the project at Chankom. We are currently receiving funding from an international agency to complete stage two at this same location. We expect that the project at Chankom will be concluded by December of 2017. On the other hand, we just started the execution of stage one at the community-UHC-Nuevo León with funding from a Mexican federal agency. We expect to conclude the project at this site by September of 2018.

Discussion

An Alternative Approach

This project represents an alternative approach for developing csCBR programs for low-income communities. This alternative considers both the research and the practice involved for the creation and execution of this type of program, and follows a participatory research approach. The main theoretical assumptions that give foundation to this project are: (1) a partnership between the community and academia is ideal, because they have different, noncompetitive, but yet complementary agendas (communities are more interested in their social development and well-being, while academia is more interested in producing and disseminating knowledge); (2) it is possible to construct new knowledge from the fusion of horizons between the community and academia; (3) reaching agreement through communicative practices will result in actions that promote social change; and (4) it is possible to build, understand, and evaluate complex multilevel interventions through the application of quantitative and qualitative methods.

The primary motivation behind this project is a need for interventions directed to reducing musculoskeletal-related disability identified by health professionals and academics. This need was informed by diverse experiences of professionals and researchers interacting with disabled people in low-income communities. Therefore, this project is the result of a genuine real life concern about the lack of social justice present in the lives of people living with musculoskeletal diseases in low socioeconomic geographic locations.

Historically, the development of CBR programs within developed and developing countries have presented some issues. These issues include the “one size fits all” strategy that is used to build such programs without considering the gap between what is needed and what is available within a community [77]. This is linked to the fact that many CBR programs have tried to import the model of “hospital rehabilitation care” directly to the community [31], resulting in “disempowering” practices [78] that aim to empower individuals without addressing “social inequalities” [79].

Our approach to csCBR program development acknowledges such problems and tries to address them through the application of a mixed method program that is “cognizant, appreciative, and inclusive of local sociopolitical realities, resources, and needs” [42]. This means that each community has to be considered as a unique entity and a general approach to build csCBR programs should incorporate efforts for adaptation to local contexts. In addition, we are proposing a grass-roots approach through the CBPR strategy. Instead of empowering individuals, this approach will aim to redistribute power, equalizing it between members of the community and academia. We believe that this strategy will counteract the inequality produced by the “charity model” [25,26] adopted by the welfare state of Mexico. In other words, instead of using the CBR program as a “band-aid” approach for solving immediate community problems [25], we are trying to promote the creation of democratic actions towards social change.

Projects of this nature will always be at risk of generating power imbalances between the members of the partnership and between the partnership and the powerful allies. This is why we are incorporating a real transdisciplinary collaboration, which involves the community and representatives from the health and social sciences. The work performed by the social scientist(s) within each stage of the project will help to disclose power imbalances, induce reflection about them, and remediate power differentials over time. This will also help to give a sense of ownership of the CBR program to all participants within the partnership and to make the collaboration with powerful allies more efficient.

Another substantial issue, registered during the development of CBR programs, is the lack of proper research and evaluation of the effects that these programs have on the disablement process within communities [41]. It is evident that evaluating these types of complex interventions is conceptually challenging. Using the traditional randomized controlled trial (RCT) approach is not feasible because of its lack of in-depth examination of the social, cultural, and organizational factors that could influence outcomes [80]. In addition, it is almost impossible to use randomization procedures within the real life situations in which CBR programs are implemented [80]. Finally, the information gathered through an RCT does not allow the capturing of the interactions between the individuals and their social and physical environments [80].

Our approach to the problem of evaluating CBR programs is to incorporate mixed methods research, in which both qualitative and quantitative methods are executed either in sequence or in parallel [42]. This implies the execution of quantitative and qualitative techniques, each one producing results that either will inform one another or will converge on a complete explanation of what is researched [73].

We opted for an ethnographic approach, due to our need for understanding the knowledge, values, and emotions towards musculoskeletal disability of people living in low-income communities within their natural settings. On the other hand, we are taking a quantitative prospective and observational approach, which will allow the use of powerful statistic tools such as multilevel analysis [81]. In addition, we are considering executing some cost-effectiveness analyses to inform the policy arena. However, at this point we would rather wait until the partnerships are well established to make decisions on how to proceed about cost analyses in the project.

Differences Between Communities

There are important differences between the community-UHC-Nuevo León and the community of Chankom. These differences have methodological and organizational implications. Nuevo León's community is 100% urban, while the Mayan community of Chankom is completely rural. This situation influences the type and consequences of existing disabling situations within these communities. The community-UHC-Nuevo León is immersed in one of the most violent Mexican States, while Chankom is situated in the least violent state of Mexico, Yucatan. This could have many repercussions on the feasibility of conducting real ethnographic work in Nuevo León because of the need for the researcher to live there for a period of time. A solution could be to locate and involve local social scientists in that area. In addition, there are important differences between communities in relation to size and spoken language. Chankom is a small indigenous community with little more than 4000 individuals who mostly speak Mayan; meanwhile, community-UHC-Nuevo León has more than 140,000 Spanish-speaking individuals. This will require constant translation efforts and the use of a significant amount of human resources. Differences between our target communities will allow us to compare between sites, advancing our understanding of the methodology required to conduct this type of project.

Another important difference between the sites involved in this project relates to the status of their local health structures and community organization development. The community-UHC-Nuevo León has a strong local primary health care system embedded in a well organized community. Whereas, there is no local health care system in Chankom and the community is poorly organized to confront their health problems. Consequently, in the community-UHC-Nuevo León we will include and share power with the community through collaboration with local health providers and community leaders since the first stage (situational analysis) of the project, which is the traditional CBPR approach. However, in Chankom we are taking a modified CBPR approach in the sense that the situational analysis will be conducted as a project driven by people from outside the community. This strategy aims to use the initial research efforts and results to motivate community organization, which will facilitate the establishment of an authentic partnership for the conduction of the next stages of the project. Chankom's situation exemplifies the difficulties encountered by trying to apply an approach developed in more organized communities to a community where organization for solving health issues is nonexistent, as are the majority of poor rural communities in Mexico.

Conclusions

In conclusion, this project is intended to move forward the methodology for the development of csCBR programs in low-income communities. These programs will contribute to community development of these Mexican socially marginalized areas and will cover the need to receive adequate health care for people living with musculoskeletal diseases at these locations.

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Authors' Contributions

ALS took the lead role to conceptualize, design, draft, and approved the final version of this protocol. JR, IPB, JL, SW, MW, JRA, JAN, DOR, RMV, and RBT contributed ideas for conceptualization and design of the protocol.

Conflicts of Interest

None declared.

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Abbreviations

6MWT: 6-minute walk test
CBR: community-based rehabilitation
CBPR: community-based participatory research
community-UHC-Nuevo León: community served by the University Health Center of Nuevo León, Monterrey, México
COPCORD: Community Oriented Program for the Control of Rheumatic Diseases
csCBR: culturally sensitive community based rehabilitation
FDT: functional dexterity test
GLADERPO: Grupo Latinoamericano para el Estudio de Enfermedades Reumaticas en Poblaciones de Origen Grupo (Latin American Group for the Study of Rheumatic Conditions in Indigenous People)
HAQ-DI: Health Assessment Questionnaire Disability Index
QoL: quality of life
RCT: randomized controlled trial
UHC-Nuevo León: University Health Center of Nuevo León, Monterrey, México
WHO: World Health Organization
WHODAS 2.0: WHO Disability Assessment Schedule 2

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Protocol

Development of a Model of Care for Rehabilitation of People Living With HIV in a Semirural Setting in South Africa

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Abstract

Background: Human immunodeficiency virus continues to challenge health care professionals even after the rollout of antiretroviral therapy. South Africa, among the worst affected countries in the world by the pandemic, has seen the effect of people living longer but facing disabling effects of both the virus and the associated impairments of the antiretroviral therapy. Rehabilitation within the evolving context of the disease has changed its focus from the impairment of the individual to the participation restriction within a person's daily life. Offering a continuum of coordinated, multilevel, multidiscipline, evidence-based rehabilitation within health care will promote its prominence in health care structures.

Objective: This study aims to develop a model of care within a health care structure using a semi-rural African setting as an example.

Methods: The study will employ mixed methods using a Learning in Action Approach into the rehabilitation of people living with HIV (PLHIV) at the study setting. The Delphi technique, a multistage consensus method, will be used to obtain feedback from a number of local experts relevant for the field of rehabilitation of people living with HIV. The study will also involve various stakeholders such as the multidisciplinary health care team (doctors, physiotherapists, occupational therapists, dieticians, speech and language therapists, social workers, midlevel workers, community health care workers); department of health representative(s); site affiliated nongovernmental organization representative(s); and service users at the study setting.

Results: Once a proposed model of care is derived, the model will be assessed for rigour and piloted at the study setting.

Conclusions: The development of a model of care in rehabilitation for PLHIV in a health care setting is aimed to provide an example of a continuum of coordinated service throughout the disease trajectory. The assumption is that the burden on the health care system will be curbed and the projected benefit for all stakeholders will promote a sort after service delivery in rehabilitation of people living with HIV.

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KEYWORDS

model of care; rehabilitation; HIV; disability; physiotherapist; Delphi technique

Introduction

Background

Human immunodeficiency virus (HIV) and the multiple effects on people living with the virus is an unequivocal health crisis facing the world at large. Sub-Saharan Africa remains the region worst affected [1]. South Africa is a middle income country at the tip of the continent that is most affected by the HIV epidemic [2]. The growing epidemic coupled with resource-poor settings in South Africa poses a challenge to the health care systems and their provision of a coordinated, multifaceted, and collaborated service from the national to the district level [2,3]. Rehabilitation is essential in facilitating patients' functional independence and has a major role in the patient flow across the health care continuum. The disabling effects of HIV together with the associated disablements of antiretroviral therapy (ART) [4,5] demands that the team of health care professionals who are involved in rehabilitation offer a range of services in managing challenges encountered by people living with HIV through a comprehensive, coordinated, and collaborated program throughout the disease trajectory [6].

A model of care in rehabilitation has the prospect to reshape service delivery, patient outcomes, efficiencies, and collaboration with health care providers across the health system. A model of care is defined as:

> *A multifaceted concept, which broadly defines the way in which health care is delivered including the values and principles; the roles and structures; and the care management and referral processes. Where possible the elements of a model of care should be based on best practice evidence and defined standards and provide structure for the delivery of health services and a framework for subsequent evaluation of care. [7]*

The impact of a model of care on the facilitative process between strategic directions for a health system to the delivery of care at local rehabilitation level is the goal within any strategic initiative [7].

The dearth of literature and lack of models of care to roll out rehabilitation for people living with HIV is astounding even though HIV and acquired immune deficiency syndrome (AIDS) are endemic in Southern Africa and therefore pose new issues to health and rehabilitation professionals in the region. Well-resourced countries have some approaches on the management of the disability in the context of HIV [8,9] However epidemic countries are still lacking such an approach and this does not only influence individual livelihoods related to disability adjusted life years but also adherence at larger scale. Thus rehabilitation needs to be integrated into the response to HIV and AIDS. The question is now not so much related to the "if" but "how" this can be done in resource poor settings such as South Africa. What model of care would address the rehabilitation needs of people living with HIV in the chosen semirural setting? Would the proposed model of care influence the rehabilitation practice at the above setting?

The aim of this study is to develop a model of care for people living with HIV in order to achieve the desired rehabilitation outcomes within a health care setting in the chosen semirural area. The objectives are to:

1. Review the existing literature and current models of care in rehabilitation;
2. Explore the perceptions of all stakeholders, that is, the multidisciplinary health care team (doctors, physiotherapists, occupational therapists, dieticians, speech and language therapists, social workers, midlevel workers, community health care workers), department of health representative(s), site affiliated nongovernmental organization (NGO) representative(s) and service users (people living with HIV receiving rehabilitation);
3. Identify criteria to be considered for the inclusion in the proposed model;
4. Develop the proposed model of care in collaboration with experts;
5. Appraise the methodological process in which the model was developed; and
6. Pilot the model of care for rehabilitation of people living with HIV within a semirural setting.

Conceptual Framework

The research will be guided by an adapted version of the "Learning in Action" approach developed by the Athena Institute in Amsterdam [10]. The study will include several sub-studies which will logically flow into each other and be developed in cooperation with the nongovernmental community partner and the Department of Health.

Approach

The process of innovation is a highly complex social set of activities, which can be conceived as an interactive, spiral process resulting from the interactions of a number of actors [10]. This implies that not only technical, but also social, organizational, political, economic, and cultural factors influence the development of innovations and the outcomes of social or technical change thereafter. The process of any type of research that focuses on innovation is therefore important to develop useful technology and interventions. However this process is often underestimated and not sufficiently understood.

On the basis of literature study and interviews the Athena Institute in Amsterdam developed the Interactive Learning in Action (ILA) approach. The approach is grounded in the following principles: active engagement of relevant stakeholders on equal footing early in the innovation process, explicit use of experiential knowledge, and knowledge creation through mutual learning (via dialogue), coalition building, and facilitated process with an emergent design. The overall process is a dynamic and cyclic process of activities, in which tentative results are tested and refined in practice in an interactive way. The ILA method comprises of five phases: (1) preparation and exploration; (2) in-depth study of problems, needs, and visions of each stakeholder group separately; (3) integration of different perspectives of stakeholder groups through mutual learning; (4) prioritization and agenda setting; and (5) implementation through reflexive cycles of planning, action, observation,

reflection, and re-planning [10]. Essential for the successful execution of the approach is a well-filled tool box of methods and tools for different functions, which can be adapted to the local context and dynamics. A combined use of qualitative and quantitative methods enables the collection and comparison of a large diversity of perspectives, and is therefore preferred.

The phased activities structured along the lines of the transdisciplinary ILA research approach allows a process of integration. In practice, the process is cyclical and dialogical; for example, the output of one stakeholder group forms the input for another group, so that information gets extensive deliberation and rigorous redefinition to a point of being widely understood and acknowledged as relevant for practical use. [10]

Each substudy of this project will be integrated in the ILA approach and focus on different levels that can influence the development of a model of care in rehabilitation to address the disabling effects of HIV in a hyperepidemic country.

Methods

Overview

The study design is a mixed methods study combining both qualitative and quantitative methods. Qualitative methods will provide rich contextual information to inform the researcher on the subjective reality experienced by participants. The quantitative methods will be employed to seek consensus and rigour in decision making and implementation of the study using the Delphi technique. Adopting this approach will lead to the convergence of expert opinion in developing a model of care where none previously existed [11]. It will allow for facilitation from international evidence based and expert informed forum to a nationally guided and local level implementation of a model of care within a rehabilitation framework.

Ethical Considerations

The study protocol received full ethical clearance from The University of KwaZulu-Natal, South Africa (HSS/1319/012D).

Setting

The study setting is a 200-bed, level one district hospital, situated on the outskirts of the suburb of Durban, in the Mariannhill Mission Complex of the province of KwaZulu-Natal, South Africa. St Mary's Hospital provides a service for 4500 people living with HIV. It is estimated that more than 250,000 people (33%) living in the St Mary's Hospital catchment area are HIV-positive. Due to the dramatic increase in the numbers of patients suffering from AIDS-related illnesses, St Mary's has focused its attention on improving and increasing its capacity to render holistic health care to HIV/AIDS patients and their families through various intervention programmes [4].

Recruitment and Selection of Participants

Participants will include stakeholders relevant to a multidisciplinary health care team (doctors, physiotherapists, occupational therapists, dieticians, speech and language therapists, social workers, midlevel workers, community health care workers), department of health representative(s), site affiliated NGO representative(s) and service users (people living with HIV receiving rehabilitation). The Delphi technique will involve experts within the physiotherapy profession with experience in HIV and rehabilitation will comprise an additional part of the panel for this consensus technique in the consequent rounds.

Data Analysis

The researcher will adopt the methodological approach of Van Manen by turning to the "nature of the lived experience, existential investigation, phenomenological reflection and conclude by phenomenological writing" [12], in analyzing the qualitative data. The Delphi multiple round questionnaires will be analyzed for level of agreement using Cronbach alpha for level of internal consistency against *a priori* limits. [13].

Results

The study has been initiated by exploring the perceptions of all stakeholders, that is, the multidisciplinary health care team (doctors, physiotherapists, occupational therapists, dieticians, speech and language therapists, social workers, midlevel workers, community health care workers), department of health representative(s), site affiliated NGO representative(s) and service users (people living with HIV receiving rehabilitation). A focus group discussion was held at the study site and the results are currently being integrated into a Delphi questionnaire.

Discussion

There is a paucity of studies in the literature addressing a model of care in the rehabilitation of people living with HIV. This study proposes the development and evaluation of a model of care using the ILA conceptual framework developed by the Athena institute which is a multilevel ILA framework incorporating a socioecological design focusing on an individual to policy development which will guide the structuring of the new "model of care in rehabilitation" within the study setting. A model of care for the rehabilitation of people living with HIV is novel and using sound theoretical discourse and evaluation will allow it to evolve from a single site study into advocating a national model to curb the burden on rehabilitation offered to people living with HIV. The study will draw on the convergence of expert opinion within the field of rehabilitation and hence provide sound theoretical judgement and critique in its development. It will allow researchers the platform to amalgamate and facilitate international evidence and expert opinion into a nationally guided and locally implemented model of care within a rehabilitation framework.

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Conflicts of Interest

None declared.

Multimedia Appendix 1

Grant award letter.

[PDF File (Adobe PDF File), 90KB - resprot_v3i4e68_app1.pdf]

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Abbreviations

AIDS: acquired immune deficiency syndrome
ART: antiretroviral therapy
HIV: human immunodeficiency virus
ILA: Interactive Learning in Action
NGO: nongovernmental organization

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Protocol

Increasing User Involvement in Health Care and Health Research Simultaneously: A Proto-Protocol for "Person-as-Researcher" and Online Decision Support Tools

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Abstract

Background: User involvement is appearing increasingly on policy agendas in many countries, with a variety of proposals for facilitating it. The belief is that it will produce better health for individuals and community, as well as demonstrate greater respect for the basic principles of autonomy and democracy.

Objective: Our Web-based project aims to increase involvement in health care and health research and is presented in the form of an umbrella protocol for a set of project-specific protocols. We conceptualize the person as a researcher engaged in a continual, living, informal “n-of-1”-type study of the effects of different actions and interventions on their health, including those implying contact with health care services. We see their research as primarily carried out in order to make better decisions for themselves, but they can offer to contribute the results to the wider population. We see the efforts of the "person-as-researcher" as contributing to the total amount of research undertaken in the community, with research not being confined to that undertaken by professional researchers and institutions. This view is fundamentally compatible with both the emancipatory and conventional approaches to increased user involvement, though somewhat more aligned with the former.

Methods: Our online decision support tools, delivered directly to the person in the community and openly accessible, are to be seen as research resources. They will take the form of interactive decision aids for a variety of specific health conditions, as well as a generic one that supports all health and health care decisions through its focus on key aspects of decision quality. We present a high-level protocol for the condition-specific studies that will implement our approach, organized within the Populations, Interventions, Comparators, Outcomes, Timings, and Settings (PICOTS) framework.

Results: Our underlying hypothesis concerns the person-as-researcher who is equipped with a prescriptive, transparent, expected value-based opinion—an opinion that combines their criterion importance weights with the Best Estimates Available Now for how well each of the available options performs on each of those outcomes. The hypothesis is that this person-as-researcher is more likely to be able to position themselves as an active participant in a clinical encounter, if they wish, than someone who has engaged with a descriptive decision aid that attempts to work with their existing cognitive processes and stresses the importance of information. The precise way this hypothesis is tested will be setting-specific and condition-specific and will be spelled out in the individual project protocols.

Conclusions: Decision resources that provide fast access to the results of slower thinking can provide the stimulus that many individuals need to take a more involved role in their own health. Our project, advanced simply as one approach to increased user involvement, is designed to make progress in the short term with minimal resources and to do so at the point of decision need, when motivation is highest. Some basic distinctions, such as those between science and non-science, research and practice,

community and individual, and lay and professional become somewhat blurred and may need to be rethought in light of this approach.

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KEYWORDS

user involvement; decision support; patient empowerment; Internet

Introduction

User involvement is appearing increasingly on the policy and action agendas of health care providers and researchers in many countries. Both "user" and "involvement" are terms broad enough to encompass a wide variety of interpretations [1-3] and to evoke a variety of proposals for how involvement can be encouraged, facilitated, and increased, regardless of interpretation. The belief is that user involvement will produce better health consequences for individual and community and will demonstrate greater respect for the basic principles of autonomy and democracy.

In discussing obstacles to such increased user involvement, the need to tackle professional attitudes, institutional barriers, and silo borders must also be emphasized [4-7]. However, some of the most fundamental barriers and borders remain largely untouched and beyond questioning, except by some at the margins of the discourse.

In our project to increase the involvement of persons in health care and health research, we find four fundamental distinctions that are problematic: (1) science and non-science, (2) research and practice, (3) group and individual, and (4) professional and lay. The four pairs are linked insofar as scientific research occurs overwhelmingly at the public group level, while professional practice, either at the individual or community level, is non-scientific. We use non-scientific in the sense that the actual application of scientifically established evidence can never be validated by the standards of science, let alone the application of beliefs or judgments. The claim that practice is evidence-based or science-based confirms, rather than contradicts, this.

Against the background of the revolution in electronic communications and computer competencies (providing widespread online access) and informatics and information storage (generating large amounts of accessible big data), we see our project, outlined here in the form of an umbrella protocol, as an addition to the variety of technologies available to optimize user involvement. But it represents a challenge to the systemic dichotomies above.

All four of the above distinctions are implicit in the activities of INVOLVE in the United Kingdom, an excellent example of an attempt to increase user involvement in health and health care *research*, in contrast to parallel attempts to increase user (ie, patient) involvement in health care *practice.* INVOLVE is a national advisory group that supports greater public involvement in the National Health Service (NHS), public health, and social care research. It is funded by and is part of the National Institute of Health Research (NIHR), which is in turn funded by the Department of Health and is tasked with sharing knowledge and learning on public involvement in research.

INVOLVE defines the public as "patients and potential patients; people who use health and social services; informal carers; parents/guardians; disabled people; members of the public who are potential recipients of health promotion programmes, public health programs and social service interventions; and organizations that represent people who use services". Public involvement in research is conceptualized as "doing research 'with' or 'by' the public, rather than 'to', 'about' or 'for' the public". INVOLVE distinguishes between three main levels of public involvement: (1) consultation (where researchers seek the views of the public on key aspects of the research), (2) collaboration (an ongoing partnership between researchers and the public throughout the research process), and (3) "publicly led" (where the public designs and undertakes the research and where researchers are invited to participate only at the invitation of the public) [8].

The split between scientist/researcher, practitioner/professional, and lay/public is clear throughout INVOLVE's descriptions but nowhere more clearly than in the final point. We see it as significant that INVOLVE has chosen to use the collective term "the public", rather than the individual term "the person", even though the former is then defined almost exclusively in terms of the latter.

Among the other instantiations of user involvement, "user *controlled* research" is a clear example of a publicly led activity, but it has political ambitions well beyond that envisaged by INVOLVE [9] (see Textbox 1).

Textbox 1. User controlled research quoted from INVOLVE [9].

-The main aim of such research is seen as liberatory; supporting the empowerment of research participants and the achievement of change in line with service users' rights and self-defined needs and interests. Such user controlled research has generally been based on:

-social rather than medicalized individual approaches and understandings;

-the rejection of positivist claims to "objectivity";

-and a commitment to personal, social and political change.

The concept of control in research is not a simple one. It may be defined in different ways and open to different interpretations. Service users and their movements, however, have identified user control as the defining characteristic of research which advances user knowledge, rights, and interests.

Community-based participatory research is less radical and more in accord with the collaborative category of INVOLVE in that it promotes a specific two-way flow of information within the research group: researchers provide information and tools to enable community members to carry out research and take action, and community members share their expert knowledge and local meanings with researchers to achieve mutual knowledge and solutions to practical problems [10,11].

Within the status quo, three types of reasons are typically given for involving users in research [12]:

> *Public involvement in health research is underpinned by epistemological, moralistic and consequentialist arguments. The epistemological argument states that health research can benefit from the experiential knowledge and personal insights of patients, carers and service users. The moralistic argument states that the public have a right to be involved in any publicly funded research that may impact on their health status or the services that they receive. Finally, the consequentialist argument states that public involvement helps to improve the quality, relevance and impact of health research.*

We suggest that a second consequentialist argument is missing from this list, particularly relevant within the setting of person-centered care [13]. In the Web-based project introduced here, we conceptualize the person as a researcher who is engaged in a continual, living, informal "n-of-1"-type study [14] of the effects of different actions and interventions on their own health, including those that imply contact with health care services. We see their research as primarily carried out in order to make better decisions for themselves, but they may offer to contribute the results to the wider population, either because it could eventually lead to better, or better-evaluated, interventions for themselves or because it could contribute to some wider public health goal or the good of others.

Within the conceptualization of person-as-researcher, those who lack the capability to function as effective researchers should be supported in their efforts to achieve that capability [15] through measures to increase health decision literacy and numeracy, especially in disadvantaged populations [16]. While we agree wholeheartedly with this principle, we note that questions of how far this support should go and at what resource cost must be part of the overall discussion of allocating scarce resources within a community, including those given to formal research. Without this reality check, all recommendations within the "capabilities" discourse remain ethically impressive but practically empty. Our project is designed to make some progress in this direction possible in the short term with minimal resources and to do so at the point of decision need, when motivation is highest.

Methods

Overview

Our online decision support tools, delivered directly to the person in the community and openly accessible, are to be regarded as "research resources". The tools take the form of interactive decision aids for a variety of specific health conditions, as well as a generic one that aims to support all health and health care decisions through its focus on key aspects of decision quality.

The tools focus directly on the person-as-researcher's fundamental question, "What should I do?" This requires answers to the two subordinate questions: "What should I believe?" and "What do I prefer?" They generate an *opinion* that integrates a set of beliefs, in the form of the Best Estimates Available Now (BEANs) for the performance of the relevant options on criteria that matter to the person, with their preferences, expressed as relative importance weights for those criteria. The integration, by a simple and transparent expected value calculation, produces a set of scores for each option that constitute the opinion produced by the process—nothing more and nothing less.

For some criteria, the person is themselves the expert source of the BEANs, since they measure the impact of options on their personal life. The difficulty, burden, or bother associated with administration routes for medications or journeys to provider facilities are good illustrations of where different individuals may make very different BEAN assessments. All persons-as-researchers contribute their individual preferences to the opinion as criterion importance weights.

Many who consult the tools in the course of their research will be satisfied that they have received a personalized opinion for their own private use. But they can offer to contribute the results of their n-of-1 research to an n-of-n database, by registering with the site by named email and declaring any conflict of interest. Their name will appear in any publication based on the aggregation of the individual results, though personal results will never be displayed. They receive feedback as part of the research team.

It is vital to be absolutely clear on one fundamental principle: whether or not the person is assigned, or accords themselves, the status of patient in some other setting, they are involved in our project as a researcher and only as a researcher. And we repeat that this approach is proposed as *one* method to be included in the portfolio of interventions needed to meet the very broad target of increased user involvement in a heterogeneous community.

From this point on the paper takes the form of an umbrella protocol for the condition-specific studies that will implement our approach. It is therefore organized using the Populations, Interventions, Comparators, Outcomes, Timings, and Settings (PICOTS) framework [17].

Populations

Our population consists of individuals researching their personal health using a more or less formal n-of-1 methodology to help decide among different health-related interventions and actions. They regard themselves as interacting with health care professionals and institutions as an individual researcher, even though they are customarily assigned the status of patient. Individuals who wish to see themselves purely as patients are advised that they may find our resources, designed to support the individual's research for better decision making,

inappropriate or unhelpful. But we hope they will proceed, subject to confirming acknowledgment of being seen in a researcher role. Those who wish to see themselves mainly, or exclusively, as patients will be well catered for by patient-centered shared decision making [18].

The focus is solely on research for better decision making about the individual's care. There is increasing interest in user involvement in relation to community-level activities, such as the development, prioritization, and delivery of health care services; the evaluation of specific interventions in Health Technology Assessments; and the determination of reimbursability for drugs and devices [1]. These are outside the scope of our project, though the approach we suggest is modifiable to this type of policy decision.

Members of the community are entitled to adopt whatever position they wish in relation to their individual interactions with health care professionals and institutions. That includes their interactions involving decision making, subject to any legal requirements, including giving informed consent. Our decision resources are, however, designed explicitly for those who wish to be able to involve themselves in clinical decision making as persons who are empowered (emancipated, enabled, armed) by their prior research. They are also intended for those who wish to keep open such positioning as an option, even if it may not eventually be exercised.

Researching one of our relevant tools will yield an opinion, based on principles that they have accepted (for their research purposes) and inputs they have supplied. We assume that the person opts into obtaining the opinion as part of the research basis for their decision involvement and emphasize that they are free to reject its content or use it in any way they wish in any subsequent decision communication with a clinician. "Clinician" should be interpreted throughout to include nurses, other health professionals, and clinical teams. "Person" should be interpreted to include the person-defined significant others and any legal guardian or proxy.

Interventions

Condition Decision-Specific Aids

Our condition decision-specific aids (eg, Should I have a prostate-specific antigen [PSA] screening test for prostate cancer? What treatment is there for my osteoarthritis?) have several characteristics that distinguish them from most other decision support tools [19,20]. We believe it is these features that carry the potential to increase user involvement, especially for the population defined above and in relation to the specified type of involvement.

While the increased scientific research on values and preferences needed for health decisions [21] proceeds, along with that on information and knowledge, clinical decisions are being made second by second. It would be wrong to say that much of the formal research being undertaken is "fiddling", though increasing concern with waste in research suggests some of it is, and even that many of the results will eventually be proven wrong [22-24]. Metaphorically, Rome is smoldering while academics are learning, and we agree with Wears that "Nothing can be gained by further perseveration in asking why clinicians fail to adopt research recommendations. Progress may come from asking, instead, why research is failing to provide useful answers to questions important to clinicians" [25]. More importantly, we should be asking questions that are important to persons-as-researchers.

As a result, and as part of our work to improve decision quality in person-centered care, we publicly offer, as research resources, decision support tools that do not require answers to many of the fundamental questions being pursued in scientific research. This is in contrast to most of the decision aids and guidelines produced within both the evidence-based and shared decision-making philosophies, which emphasize current uncertainties, ignorance, and the need for caution. We believe vague urgings to "be cautious" are unproductive, unless accompanied by some operational guidance on *how* to be cautious, given a decision is to be made *now*. We therefore make our offers on the basis that the underlying theory and principles of the aids, as well as the nature and provenance of their empirical inputs, are made clear before any engagement with them (or buy-in) is possible. The user is required to have read and accepted the contents before proceeding. We therefore assume that they are making an informed *meta*-decision about whether to engage with the aid before any further involvement, even as a researcher. An involvement strategy that proceeds without this sort of high-level consent goes beyond "persuasion-as-simply-making-available" into covert nudging at best and coercive manipulation of choice at worst. It is ethically questionable [26-28].

The aids produce an opinion based on a prescriptive model for decision making in the form of Multi-Criteria Decision Analysis (MCDA). The opinion is "dually personalized" as it consists of the scores produced by combining (in an expected value calculation) the person's percentage importance weights for the criteria important to them with the BEANs for the personalized performance of each option on each criterion. The aids make absolutely no claim to be descriptively based in human decision behavior [29]. In fact, in key respects, especially their numerical format and expected value basis, their descriptive inadequacies are a necessary condition of their having something new and important to offer [30]. The aids are presented with as much transparency as possible, in order for the person to be clear about the principles underlying the opinion that emerges. We emphasize that they can reject the opinion of the aid as a contribution to their research, having generated it, but advise that they should consider not even engaging with it if they disagree with the bases spelled out upfront.

While we refer to "preferences", our precise term is "importance weights". As with most other terms in this area, debate surrounds its meaning. We define importance weights simply as the normalized responses of a respondent asked "How important is [each criterion] to you on an 11-point scale ranging from 0='of no importance' to 10='of extreme importance'?". After the responses are transformed into weights adding to 100% by normalization, the respondent has the opportunity to use the cursor or touch to modify the bar-length representations presented on the screen. We regard this elicitation procedure as the only one that is practical, in comparison to the more complex, normatively appealing procedures such as standard

gambles, time trade-offs, and swing weights, which we have tried and found operationally lacking [19]. We do not take any position on whether these importance weights meet anybody's normative requirements for constituting "utilities". The key point is again to make clear to the respondent that it is their importance weights, so defined, that are entered into the personalized opinion that the aid will produce for them as part of their research.

In regard to the performance rates for options on criteria, our tools are *not* designed primarily as information aids. They are therefore clearly different from most other aids that assume a better decision must be an informed decision. We do provide links to high-quality sources of information so that the person-as-researcher can opt in to them if they choose. But it is made clear that our primary aim is to provide information in the form of the BEANs for the performance of each option on each criterion. These are updated within a "living" philosophy [31] and reject any generic value-judgment based threshold (eg, $P<.05$) for what is usable in clinical decision making. In the absence of robust evidence, they may be best elicited by expert-based elicitation. The BEANs entered into the individual's aid are personalized as much as possible on the basis of self-reported characteristics. Opt-in pop-ups provide the provenance of the BEANs, or links to their sources, and the person-as-researcher is free to follow these as further clues to trustworthiness. Why do we not regard these as vital to consult in order to benefit from the aids? Because we are aiming solely to provide an opinion based on an expected value calculation that synthesizes the BEANs with the person's importance weights. Given this purpose, there is no need to communicate about the size or quality of the detailed BEANs in the way typically envisaged by those who see "risk communication" as a central task in informed decision support. Achieving success in this task is difficult [32], perhaps not surprising in the light of the failures of the educational and socialization systems to produce a health literate and numerate population. The only information our person-as-researcher needs to acquire is what the aid will provide and its bases—and what it does *not* offer.

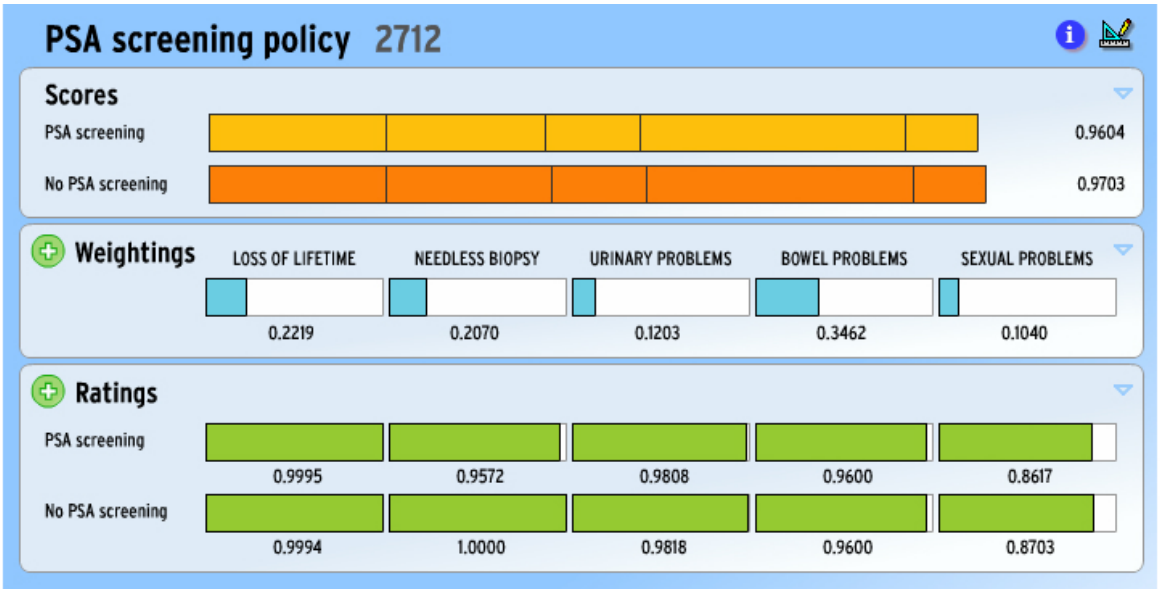
However, there is an important exception. The person-as-researcher does have an important role in supplying, at the point of decision, the BEANs for criteria where they are the expert. This is notably the case regarding the impact of testing and/or treatment on the individual as a person or party to a relationship. The rating of the burden or bother associated with, for example, different modes of treatment delivery (eg, oral, topical, subcutaneous injection, intravenous infusion; home, clinic, hospital) will vary with an individual's workloads and capacities [33]. Personalized elicitation of the BEANs for such criteria is therefore appropriate—not the use of group averages such as those produced by discrete choice experiments. Note that this rating role of the user is conceptually completely different from the role they play in assigning an importance weighting to such criteria, relative to all the others.

Uncertainty is dealt with by offering quality-weighted and unweighted opinions. We make clear that the quality adjustments in the former represent, no more and no less, the judgments of the quality of the BEANs made by the team responsible for their production.

Our aids, such as "Should I have a PSA screening test for prostate cancer?" (Figure 1), are the product of teams of named health professionals, including clinicians. But we stress that the opinion emerging is not offered as, and should not be interpreted as, a medical opinion, legally speaking.

Most of the key requirements for accessibility, usability, and functionality of patient-centered decision support, whether they come in the form of computer-based decision aids or traditional professional interaction, apply equally to the design of aids to be presented as research resources [34-36]. Nevertheless, the re-conceptualization from patient to person-as-researcher does have major implications in the tone of address and register adopted. Most importantly, our decision aids should not be seen in any way as providing care, or as a way of delivering better care. Instead, they are intended simply as an optional resource available in the person's own pursuit of the sources of better care. However, they also provide a way that users can add the results of their engagement to those of others, if they choose.

Figure 1. Example of PSA decision aid screen.



A Generic Decision Aid: MyDecisionQuality

User involvement is for a purpose, and our central aim is to improve decision quality. A measure of effectiveness in this regard is obviously needed.

MyDecisionQuality (MDQ) is a dually personalized decision quality instrument based (as are our condition decision-specific aids) on MCDA) [37]. The assessor (eg, the person) is responsible not only for (1) weighting the criteria of decision quality in terms of their relative importance, but also (2) rating the quality of a decision just made on the criteria. MDQ is generic in the sense that the criteria are phrased without reference to any particular decision or context. Information relating to the specific decision condition and setting must be provided (if at all) outside the MDQ instrument, such as in the wider condition-decision support resource where it will often be situated.

As with all implementations of the simple weighted-sum version of MCDA, MDQ combines a set of importance weights for multiple criteria with performance ratings for each option on these criteria and calculates the overall score as the expected value of these components. In the case of MDQ, the person's weightings for the eight criteria of decision quality are elicited as early as possible in the decision-making process, and their ratings on how well the decision made performed on these criteria, as soon as possible after it was made. The MDQ score, unique to the person and to the particular occasion, is shown with the partial contributions of each criterion to it displayed in segments. Its weighting and rating are highlighted when the segment is touched or the cursor is rolled over it. An example is provided in Figure 2 and an illustrative video in Multimedia Appendix 1.

Apart from serving as an outcome measure for evaluating the decision-making process, MDQ represents an aid in itself and, being generic, can be used in conjunction with any of our condition decision-specific aids. Independent of any health care context or setting, MDQ alerts the person-as-researcher to one set of criteria for a good decision and asks them to express their importance weights for them. Even if these weights are not widely different from each other—not unusual since the criteria have been included because of their importance—the explicit attention given to them has the potential to influence the remainder of their decision-making research. Having rated the decision ex post on the same criteria, the person receives a dually personalized assessment of the quality of their decision. They are also provided with insight into the priorities for future quality improvement by being shown the quality gains possible from improved rating on each criterion, weightings unchanged. For example, in Figure 2 we can inform the person of the effect on their decision quality score of improving their rating on "Importance", lowly rated at 0.3, given the relatively high weight of 0.188 they have assigned it. Achieving perfect rating on this criterion would increase their score by 0.7 x 0.188 or 0.132, equivalent to a 20% improvement. Feeding back the result of the same calculation for each of the criteria generates a personalized list of future priorities. Since the criterion "Effects" is already highly rated, it is unlikely to be high on this priority list, even though it has the same weight as Importance.

If an associated clinician completes the parallel MDQ instrument, the bases for a decomposable measure of concordance are established. A prescription for improved shared decision making in future is generated, if desired by both parties. It can help reduce the established differences in a person's preferred and perceived participation in medical decision making [38].

MDQ can also serve as a patient-reported outcome measure (PROM), when the decision is conceptualized as one of the outcomes of a decision-making process. Or alternatively, it can be seen as a patient reported experience measure (PREM), which reflects their decision-making experience [39,40].

A bonus resulting from the use of both condition decision-specific and generic aids comes in the form of the enhanced and automatic documentation of the clinical decision-making process, given that the outputs can be saved by the person-as-researcher and incorporated into their provider's and own health record/s, if desired.

Figure 2. Example of MyDecisionQuality screen.

Comparators

Apart from a few aids also based on an implementation of MCDA (notably the Analytic Hierarchy Process), the vast majority of decision support tools on offer are not designed to produce an opinion in the form of numerical scores. They aim to support the person, normally regarded as a patient, by presenting information and value clarification exercises. They are then encouraged to make up their mind by taking into account and bearing in mind the pros and cons, without being offered explicit synthesizing principle or required to engage in numerical quantification or calculation. We can capture the difference from their aids succinctly by referring to the majority as being grounded in verbal multi-criteria decision deliberation as opposed to ours in numerical MCDA. Note that one of the key contrasts is expressed here as the verbal-numerical, rather than qualitative-quantitative one. All aids of both types are necessarily concerned with quantifying of magnitudes.

Our underlying hypothesis concerns the person-as-researcher who is equipped with a prescriptive, transparent, expected value-based opinion that combines their criterion importance weights with the BEANs for how well each of the available options performs on each of those outcomes. The hypothesis is that this person-as-researcher is more likely to be able to position themselves as an active participant in a clinical encounter, if they wish, than someone who has engaged with a descriptive decision aid that attempts to work with their existing cognitive processes and stresses the importance of information. Research that opens the "black box" of the clinical encounter [41,42] is revealing less and less impact from the latter approach to decision support. Most likely this is attributable to their failure to provide the person with powerful enough ammunition to move clinicians away from their preferred consultation structure and preferred course of action, reflecting tradition, training, and time constraints. This is particularly likely to happen in the situation where the evidence is low [43].

Apart from being provisional, the opinion from our aids will always be questionable by the normative standards built into many checklists for decision support tools [44,45]. We regard the relevant comparator as an empirical one, in the form of today's clinician, and not abstract normative perfection. Experience so far shows there are many difficulties in carrying out genuinely unbiased empirical evaluations of person-centered decision aids in the clinical context—some methodological, some professional, and others legal.

Outcomes

The black box metaphor is highly relevant in relation to the question that may be uppermost in some reader's minds. What and where is the evidence of the impact of resources such as ours on any aspects of clinical decision making, notably user involvement and empowerment? A substantive, not merely rhetorical, response is to ask what and where the evidence is concerning the usual clinical decision-making process. Despite vast efforts to penetrate it, dating back to the pioneering work of Elstein [46], our aids will, by comparison, be shining white boxes.

We note with interest that clinicians and health care institutions are largely free to introduce practice changes as "quality improvements" without citing any robust evidence base or reference to peer-reviewed evaluations. In person-centered care, it is surely appropriate to acknowledge individuals have the same right in regard to their health decisions and behaviors. Using online decision resources of our type, under their explicit ground rules, falls well within our concept of the person's self-seeking quality improvement in health decision making, whether alone or in collaboration with clinicians.

Nevertheless, in the context of growing funding of research into interventions that (might) increase user involvement, serious evaluation is needed of both effectiveness and cost-effectiveness, with "multi-criteria" preceding "effectiveness" in both cases. Hence this high-level protocol, designed to set out the relevant issues. In our opinion, all user involvement interventions should be evaluated with a comparative methodology using the same empirical comparator, not a normative checklist. In other words, evaluation should be based on the same principles applied to drugs and devices. The relevant comparator will necessarily be a "usual practice" arm, and we welcome the opportunity to engage in an empirical comparison with all other proposed interventions on a "level playing field". Unfortunately, experience shows the ethical and professional barriers to this may be considerable. Authorities contemplating evaluation and resourcing of alternative user involvement strategies should therefore be aware that the position they take on professional and ethical issues may well bias the result in a particular direction. That direction is more likely to be towards

institutionalized forms of user representation and consultation than towards the more profound involvement envisioned within user controlled research, participant action research, and other emancipatory movements.

Timing

Decision time is always *now*, so our tools are developed and maintained within a living philosophy [31], especially, in relation to the performance ratings, where living evidence-based network meta-analyses will need to be complemented by expert elicitation, to improve the quality of the BEANs for many person-important criteria. Elicitation could possibly be in the form of living expertise-based network meta-analyses [47].

Settings

Our decision resources are designed to be practical for use at home in the community. This use may or may not be prior to some arranged or contemplated clinical consultation, depending on the individual person-as-researcher's wishes. Their subsequent use in the clinical setting would be subject to the clinician's agreement. Practicality in the home situation is the key to use of a resource designed to increase involvement. This will necessarily involve persons-as-researchers being allowed to make their own time and resource trade-offs in pursuing the complexity and depth offered.

Results

As implied in the Comparator and Outcomes sections of the protocol, our underlying hypothesis concerns the person-as-researcher who is equipped with a prescriptive, transparent, expected value-based opinion—an opinion that combines their criterion importance weights with the Best Estimates Available Now for how well each of the available options performs on each of those outcomes. The hypothesis is that this person-as-researcher is more likely to be able to position themselves as an active participant in a clinical encounter, if they wish, than someone who has engaged with a descriptive decision aid that attempts to work with their existing cognitive processes and stresses the importance of information. The precise way this hypothesis is tested will need to be setting-specific and condition-specific, and these details will be spelled out in the individual project protocols.

Discussion

Other Considerations

The most advanced involvement of patient representatives in health research design and activity has been in OMERACT (Outcome Measures in Rheumatology) [48]. While important effects have been achieved, especially in adding person-important criteria such as fatigue to core outcome measures, the picture is not all rosy. Some participants in meetings have felt that "Dealing with hierarchical power relations and strongly opinionated professionals was experienced as mentally challenging. A recurring barrier reported by patients was a lack of feedback on provided contributions. At times they felt that their experiential knowledge was not accepted as a valid source for scientific research, nor seen as relevant compared to the expert knowledge of professionals" [48]. While this approach is likely to become more popular and effective, it will always be confined to a small number of patients. We seek much wider involvement through the open-access resources outlined in this paper.

Clinical decision making occurs as the final "bedside" stage of most translation models of the research-into-practice process. In many ways, it is the most complex stage to understand, to assess, and to intervene. We believe the Callard model is the most appropriate one for a person-centered health care system [49]. The user, now person-as-researcher, is separately placed in the middle of the model, rather than at the end of a translation pathway, or at one point in a cyclical translational system. Consequently they have direct impact on, and input into, all stages on the forward translation continuum from "bench to bedside". In a small but significant modification to the Callard model, we suggest the person-as-researcher at the center is equipped with a decision support tool based on person-important criteria. The BEANs in their personalized resource represent the product of all necessary and practical forward translations needed at the point of decision, while the assessed quality of the BEAN for each cell constitutes the basis for backward translation to research priorities. In contrast (but not opposition) to the James Lind Alliance approach, which focuses on developing specific questions for researchers [50], priorities are indicated by the potential score gains for options from higher quality criterion ratings, given the criterion weights.

Conclusions

Even a superficial overview of recent calls for increased user involvement in health care systems reveals a complex mix of motivations and interpretations. These are reflected in the diversity of terms and interpretations for both user (client, customer, patient, person) and involvement (participation, engagement, activation, emancipation). It is not surprising, then, that many and varied approaches to increasing user involvement have been canvassed, and implemented in some cases, without serious, comparative empirical evaluation.

In the light of this, our paper has had two purposes. The first explicit aim is to offer our specific person-as-(n-of-1) researcher approach that increases the individual's involvement in health care practice and health care research simultaneously. The basis of the approach, through online interactive decision tools available as open access resources, differs significantly from most others on offer, and these differences extend to the theoretical and empirical bases of the aids. These have been described at length. The second implicit aim is to call attention to the need for careful and thorough specification, evaluation, and resourcing of programs or projects set up to achieve the broad aim of increased user involvement. Since there will be many considerations and stakeholders in play, both conceptual clarity and policy transparency make some form of multi-criteria analysis almost essential as policy decision support. A technique such as MCDA can ensure that the specifications of the options and criteria are precise and comprehensive. It will also ensure that the ratings of the options on each of the multiple criteria, which are likely to vary among stakeholders, are elicited and processed in a way that makes their provenance transparent.

Web-based decision resources such as those we produce can provide fast and efficient access to the results of slower thinking and encourage individuals to take a more involved role in their health production by viewing themselves as researchers involved in ongoing n-of-1 type studies.

Some basic distinctions, such as those between science and non-science, research and practice, community and individual, and lay and professional become somewhat blurred and will need to be rethought in the light of this approach. We encourage others to engage with us in this rethinking.

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Authors' Contributions

JD and MKK jointly developed the reconceptualization of the person as researcher and of online decision aids as a decision resource that could become an open one. JD wrote the initial draft, which MKK revised. GS's and JBN's comments were incorporated in the final version by JD. All authors approved this version.

Conflicts of Interest

Jack Dowie has a financial interest in the Annalisa software used in the Annalisa-based decision aids and the current implementation of the MyDecisionQualty instrument at the University of Sydney but does not benefit from its use there.

Multimedia Appendix 1

Video demonstration of MyDecisionQuality.

[MP4 File (MP4 Video), 923KB - resprot_v3i4e61_app1.mp4]

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Abbreviations

BEAN: best estimate available now
MCDA: multi-criteria decision analysis
MDQ: MyDecisionQuality
OMERACT: Outcome Measures in Rheumatology
PICOT: Populations, Interventions, Comparators, Outcomes, Timings, Settings
PSA: prostate-specific antigen

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Protocol

Experimental Design to Evaluate Directed Adaptive Mutation in Mammalian Cells

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Abstract

Background: We describe the experimental design for a methodological approach to determine whether directed adaptive mutation occurs in mammalian cells. Identification of directed adaptive mutation would have profound practical significance for a wide variety of biomedical problems, including disease development and resistance to treatment. In adaptive mutation, the genetic or epigenetic change is not random; instead, the presence and type of selection influences the frequency and character of the mutation event. Adaptive mutation can contribute to the evolution of microbial pathogenesis, cancer, and drug resistance, and may become a focus of novel therapeutic interventions.

Objective: Our experimental approach was designed to distinguish between 3 types of mutation: (1) random mutations that are independent of selective pressure, (2) undirected adaptive mutations that arise when selective pressure induces a general increase in the mutation rate, and (3) directed adaptive mutations that arise when selective pressure induces targeted mutations that specifically influence the adaptive response. The purpose of this report is to introduce an experimental design and describe limited pilot experiment data (not to describe a complete set of experiments); hence, it is an early report.

Methods: An experimental design based on immortalization of mouse embryonic fibroblast cells is presented that links clonal cell growth to reversal of an inactivating polyadenylation site mutation. Thus, cells exhibit growth only in the presence of both the countermutation and an inducing agent (doxycycline). The type and frequency of mutation in the presence or absence of doxycycline will be evaluated. Additional experimental approaches would determine whether the cells exhibit a generalized increase in mutation rate and/or whether the cells show altered expression of error-prone DNA polymerases or of mismatch repair proteins.

Results: We performed the initial stages of characterizing our system and have limited preliminary data from several pilot experiments. Cell growth and DNA sequence data indicate that we have identified a cell clone that exhibits several suitable characteristics, although further study is required to identify a more optimal cell clone.

Conclusions: The experimental approach is based on a quantum biological model of basis-dependent selection describing a novel mechanism of adaptive mutation. This project is currently inactive due to lack of funding. However, consistent with the objective of early reports, we describe a proposed study that has not produced publishable results, but is worthy of report because of the hypothesis, experimental design, and protocols. We outline the project's rationale and experimental design, with its strengths and weaknesses, to stimulate discussion and analysis, and lay the foundation for future studies in this field.

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KEYWORDS

quantum; adaptive mutation; mouse embryo fibroblast

Introduction

Adaptive Mutation

Random biological mutations occur independent of selection pressure. Although this likely describes most mutations, adaptive mutation may also contribute to genetic variability in changing environments. In adaptive mutation, the genetic change does not exist independent of the selective pressure; instead, the presence and type of selection influences the frequency and character of the mutation event. Evidence for adaptive mutation exists for both bacteria and yeast, and possibly for prostate cancer cells; researchers believe that adaptive mutation contributes to the evolution of microbial pathogenesis, cancer, and drug resistance, and may become a focus of novel therapeutic interventions [1-22].

This proposal evaluates the possibility of directed adaptive mutation in mammalian cells. Our objective is to distinguish between 3 types of mutation: (1) random mutations independent of selective pressure; (2) undirected adaptive mutations, which arise when selective pressure induces a general increase in the mutation rate; and (3) directed adaptive mutations, which arise when selective pressure induces targeted mutations that specifically influence the adaptive response.

A number of hypotheses have been postulated to explain undirected adaptive mutation. These include replication and recombination systems, slow repair of mismatched bases, mutagenic transcription, and gene amplification/duplication (reviewed in [17]). The most cited potential mechanism for undirected adaptive mutation is induction of a transient hypermutagenic "mutator phenotype," in which the mutation frequency is increased by up to several orders of magnitude [4,6,18]. The mutator phenotype has been invoked to explain the development of resistance to the androgen receptor antagonist bicalutamide in prostate cancer cells [16], a possible example of undirected adaptive mutation. LNCap prostate cancer cells respond to a bicalutamide challenge by upregulating expression of error-prone DNA polymerases and downregulating expression of high-fidelity DNA polymerases and mismatch repair (MMR) proteins, resulting in an increased mutation rate [16]. However, we are interested in evaluating the possibility of directed adaptive mutation, defined as matched specific mutations with associated specific environmental changes (eg, the targeted mutation of one gene to a single specific selective pressure). This hypothesized form of adaptive mutation cannot be explained through a generalized increase in mutation rate.

Basis-Dependent Selection and Quantum Biology

Ogryzko [23-26] and, more recently, Bordonaro and Ogryzko [27] proposed a quantum biological mechanism for adaptive mutation (Multimedia Appendix 1) in which superposition is context-dependent. In biological systems, context-dependence describes possible environmental conditions; thus, a change of environment represents a change in the basis (ie, basis transformation) describing a quantum mechanical system. Whether or not a quantum state is in a state of superposition (quantum coherence) is strictly dependent on the basis used to describe the system. A state that can be described as being in superposition in one basis may not be so described in another basis, and vice versa. Thus, the state of a biological system can be stable in a given environment and not affected by decoherence; however, upon a change in environment (change of basis), that same biological system can be described as being in a state of superposition. Decoherence would follow, resulting in a new set of stable, preferred states, one of which would be observed on measurement. Because there is no requirement to maintain quantum coherence for some arbitrary time period in any arbitrary environment, past criticisms [28] of quantum biology (eg, "biological systems are too warm and complex to maintain quantum coherence") are not relevant.

Quantum superposition is known to contribute to several biological processes (reviewed in [27]); however, the role of quantum effects in adaptive mutation has not been established. The hypothesis of basis-dependent selection attempts to address this deficiency through a novel model of directed adaptive mutation [27]. Thus, consider 2 cell states (A_1 and A_2, with A_2 representing a cell state characterized by a gene mutation allowing for cell growth) and consider 2 possible environmental states (B_1 and B_2, such that only B_2 allows for cell growth). Cell growth will occur only with cell state A_2 in environment B_2. A mutation that enables cell growth (A_2) would occur only (1) in an environment suitable for cell growth (B_2) and (2) only after exposure to that specific environment. Therefore, each specific microenvironment is correlated with a specific set of potential cell states (eg, cell states characterized by wild-type or mutant DNA sequences). These mutations do not occur randomly; instead, the cellular microenvironment (B_2) selects the possible spectrum of cell states (A_1 and A_2) possible in that environment. An irreversible change in the state of the cell (eg, clonal expansion) would establish state A_2 as that which is observed. If A_2 represents a mutant cell state, this process can be described as directed adaptive mutation A_2 induced by the selective pressure of environment B_2. Note that the selection process is dependent on the environment of the biological system; hence, it is basis-dependent selection leading to adaptive mutation.

It is acknowledged that adaptive mutation, including what we define here as "directed" adaptive mutation, could in each case be dealt with through theories invoking classical mechanisms, particularly when ad hoc explanations are used. However, we believe that the difference between progressive and regressive research programs, previously discussed with respect to basis-dependent selection [27], is of relevance. The hypothesis of basis-dependent selection makes testable predictions, which would be evaluated via our experimental design. Specific findings (eg, directed adaptive mutation in our system) would be better explained by quantum than by classical mechanisms, and the basis-dependent selection hypothesis leads to further testable predictions that can be evaluated by experiments

extending our approach. Future refinements in methodology may more definitively identify the fundamental nature (eg, quantum vs classical) of the mechanisms driving adaptive mutation. This would test predictions generated by the findings of the study outlined here, leading to more refined hypotheses and additional testable predictions. There is inherent value in proposing, and testing to the extent we are able, basis-dependent selection as a mechanism driving adaptive mutation. Thus, our experimental design is informed by the progressive research program suggested by previous discussions of basis-dependent selection [23-27].

Experimental Design

In our system, expression of SV40 large T antigen (TAg) is required for clonal cell growth, and expression of TAg is dependent on environmental conditions. Thus, we use an inducible system in which TAg expression and resultant clonal cell growth occurs in the presence of doxycycline. However, the TAg expression cassette is designed to contain a mutation that prevents TAg expression; therefore, unless the mutation is reversed, clonal cell growth would not occur even in the presence of doxycycline. Thus, in this system, clonal cell growth requires (1) doxycycline and (2) that the inactivating mutation is reversed by a countermutation allowing TAg expression. In the absence of doxycycline, the cells would not grow regardless of countermutation; conversely, in the absence of countermutation, treatment with doxycycline would not induce cell growth. Therefore, our prediction is that countermutation (ie, another mutation) would occur in a directed manner dependent on the appropriate environmental conditions for growth (ie, doxycycline).

This experimental model can be theoretically interpreted according to basis-dependent selection [27] as follows. In environment B_1 (no doxycycline), cell states A_1 (no countermutation) and A_2 (countermutation) cannot be distinguished because cell growth cannot occur in the absence of doxycycline. However, in environment B_2 (presence of doxycycline), the 2 cell states can be distinguished because only cell state A_2 is capable of responding to doxycycline with clonal growth. In the doxycycline-treatment environment, the 2 “A” states, which can now be distinguished, are stable, preferred states of the cell system. Therefore, in the presence of doxycycline, the superposition of cell states A_1/A_2 evolves through (1) decoherence into the 2 stable, preferred states, followed by (2) observation of clonal growth (cell state exhibiting countermutation A_2) through the irreversible process of proliferation of cells exhibiting state A_2.

Using this approach, we have devised an experimental system based on inducible and reversible cell immortalization. In this setting, mouse embryo fibroblast (MEF) cells require TAg expression for immortalization and clonal growth. MEF cells will be stably transfected with a TAg expression plasmid (Figure 1) that allows for clonal growth dependent on doxycycline-induced TAg expression. In the absence of doxycycline, cells should not express TAg or undergo clonal growth, while maintaining the ability to exhibit efficient growth (“reimmortalization”) if doxycycline is added to the growth medium [29]. The structure of the plasmid is shown in Figure 1. A bidirectional promoter allows for expression of tetracycline (Tet) repressor, neomycin-resistance gene, and green fluorescent protein (GFP) in one direction, and for expression of TAg, in a doxycycline-inducible manner, in the other direction [29]. Because these MEF cells require TAg for continuous growth, the cells must express TAg for routine cell culture and cell expansion. Therefore, a characteristic of our system is that (1) wild-type TAg expression can be used to expand the cells as needed, but (2) the inactivating mutation is introduced at the start of the experimental protocol, eliminating TAg expression when desired. The mutation utilized is in the polyadenylation (polyA) site hexanucleotide sequence for the TAg expression cassette.

The controlled pattern of TAg expression will be accomplished by inserting tandem polyA sites downstream of the TAg sequence (Figure 1). Immediately downstream of TAg is a floxed (“flanked by lox sites”) polyA cassette containing 3 wild-type SV40 (L) polyA sites. This functional polyA cassette allows for TAg expression and cell growth during routine cell culture before the start of the experiment. Further downstream is another SV40 (L) polyA site containing a mutated hexanucleotide sequence. The polyA site mutation is designed to repress 3′ end processing [30], eliminating TAg expression, and inducing cell quiescence at the start of the experiment. When the upstream wild-type polyA cassette is present, the transcripts undergo efficient 3′ end processing, allowing for TAg expression and MEF cell growth (Figure 2). However, when the wild-type polyA cassette is removed by CRE-Lox excision, only the mutated inactive polyA site (Figures 1-3) remains. The resulting defect in 3′ end processing should repress TAg expression and inhibit MEF cell growth. Clonal cell growth would occur only on reversal of the polyA site mutation, restoring efficient 3′ end processing and TAg expression in the presence of doxycycline.

Therefore, an outline of the overall experimental design for this project (Figure 1) is as follows. pRITAGKHexa and a plasmid containing a floxed hygromycin-thymidine kinase (Hygro-TK) expression cassette would be stably cotransfected into MEF cells and the cells incubated with G418, hygromycin, and doxycycline. TAg expression allows for cell growth. Removal of the floxed SV40 polyA sites from the integrated RITAGKHexa sequences, and the Hygro-TK cassette from the integrated Hygro-TK plasmid, would be achieved by CRE expression from an adenovirus (Figure 1). Thus, when cells transfected with the experimental plasmid are exposed to CRE recombinase, the floxed polyA sites are removed, leaving behind a mutant SV40 polyA site as the only polyA site for TAg expression. Cells would be taken off hygromycin and subsequently treated with ganciclovir to kill cells that did not undergo CRE-mediated excision (and still express TK). At the same time, doxycycline is removed from the media and the cells enter a potentially reversible stasis. Doxycycline would then be added back to the media; cells that retain the mutant polyA site would not efficiently express SV40 TAg and would not grow, but those cells that exhibit a further “reversion” mutation to wild-type polyA sequence would reexpress SV40 TAg and commence growth. If the latter occurs, follow-up experiments

would determine whether the mutation to wild-type polyA status was adaptive or preexisting.

Thus, the initial characterization data derived from pilot experiments clearly show that the basics of the system are working as expected, for SV40 TAg expression and cell growth (Figure 2), as well as for the presence of the correct mutant polyA sequence in the stably transfected MEF cells (Figure 3). The experimental protocols utilized to generate these preliminary data were as follows. For analysis of SV40 TAg protein expression, MEF cells stably transfected with the pRITAGKHexa and Hygro-TK plasmids were infected or mock infected with Ad-CRE adenovirus at a multiplicity of infection (MOI) of 1000. Cells exposed to Ad-CRE were treated with 50 μM ganciclovir to eliminate cells that were not infected and did not undergo CRE-Lox excision. Three days later, protein was isolated and Western blot analysis performed with anti-SV40 TAg and anti-actin antibodies. SV40 TAg is expressed before (no CRE), but not after (CRE), excision of the wild-type polyA cassette; excision leaves behind only the mutant hexanucleotide polyA site. To analyze doxycycline-dependent cell proliferation, MEF cells were plated out at 4000 cells/well in a 96-well plate either in the presence of 4 μg/mL doxycycline, 400 μg/mL G418, and 100 μg/mL hygromycin (+Doxy) or in the presence of hygromycin alone (–Doxy). Note that the neomycin-resistance gene expression is also under the control of the doxycycline-inducible promoter, so the –Doxy samples are incubated in the absence of G418 to prevent possible toxic effects of G418 from complicating the analysis of cells in the –Doxy condition. Cells were assayed with the QuickCell Proliferation kit. For doxycycline-dependent clonal cell growth and reanimation, 500 MEF clone F cells were plated in a 6-well plate in the presence of 4 μg/mL doxycycline, 400 μg/mL G418, and 100 μg/mL hygromycin or hygromycin alone. All cells were grown with a 1:1000 dilution of Fungizone. At the end of the experiment, colonies were stained with crystal violet. To analyze the DNA sequence of the mutant hexanucleotide in stably transfected MEF cells (Figure 3), genomic DNA was isolated and the relevant sequence amplified by polymerase chain reaction and sequenced (Genewiz).

To summarize, in this experimental design, MEF cells would be stably transfected with a plasmid (pRITAGKHexa) (Figure 1) expressing TAg from a doxycycline-inducible promoter, with a floxed wild-type polyA cassette and a downstream mutated polyA site (Figures 1-3). Cells would then be cotransfected with a plasmid expressing a floxed hygromycin-thymidine kinase (Hygro-TK) expression cassette; TK expression activates the prodrug ganciclovir, killing any cells retaining the Hygro-TK cassette. The purpose of ganciclovir selection in our system is to ensure that the only cells subjected to the experimental conditions are those that have successfully undergone CRE-Lox excision of the relevant floxed DNA sequences. Thus, the first steps of the main experimental protocol would be as follows. Cells would be infected with an adenovirus expressing CRE recombinase, which would remove both the wild-type polyA cassette from the integrated pRITAGKHexa plasmid and the Hygro-TK cassette from the integrated Hygro-TK plasmid. Cells would be plated out in replicates, with or without doxycycline, and with ganciclovir, which would kill any cells retaining the Hygro-TK cassette. Therefore, cells not infected with the adenovirus and that have not undergone CRE-Lox excision of floxed DNA sequences would be removed from the cell population. This is an important step because any cells not infected with CRE adenovirus would efficiently grow in the presence of doxycycline even without countermutation of the downstream polyA site. Thus, in noninfected cells, the continued presence of the floxed upstream wild-type polyA cassette would result in efficient 3′ end processing, high TAg expression, and clonal cell growth. Killing noninfected cells would eliminate the possibility of false positive results due to clonal growth of noninfected cells retaining the upstream polyA cassette. We would subsequently observe if infected cells treated with doxycycline form colonies suggesting the possibility of a reversal (countermutation) of the original polyA site mutation. Our preliminary data (Figure 4) demonstrate a lack of false positive results in 3 replicates of a pilot experiment infection.

The specific protocol utilized to generate these preliminary data (Figure 4) was as follows. A total of 150,000 MEF clone F cells were plated in a 6-well plate in the presence of 4 μg/mL doxycycline, 400 μg/mL G418, and 100 μg/mL hygromycin (F+). The next day, cells in media without the aforementioned added reagents were infected with Ad-CMV-CRE virus at a MOI of 1000 (assuming cell doubling). After 24-hr infection, cells were trypsinized and replated onto a 15-cm dish with 400 μg/mL G418 and 50 μM ganciclovir. After a further 24 hr, 4 μg/mL doxycycline was added to the media. Cells were incubated with doxycycline, G418, and ganciclovir (with regular changing of media) until the end of the experiment; at the end of the experiment (19 days postreplating), colonies were stained with crystal violet. All cells were grown with a 1:1000 dilution of Fungizone except during the 24-hr virus infection.

Figure 1. Basic strategy for pRITAGKHexa experiments. (A) Plasmid structure. (B) Outline of the experimental approach.

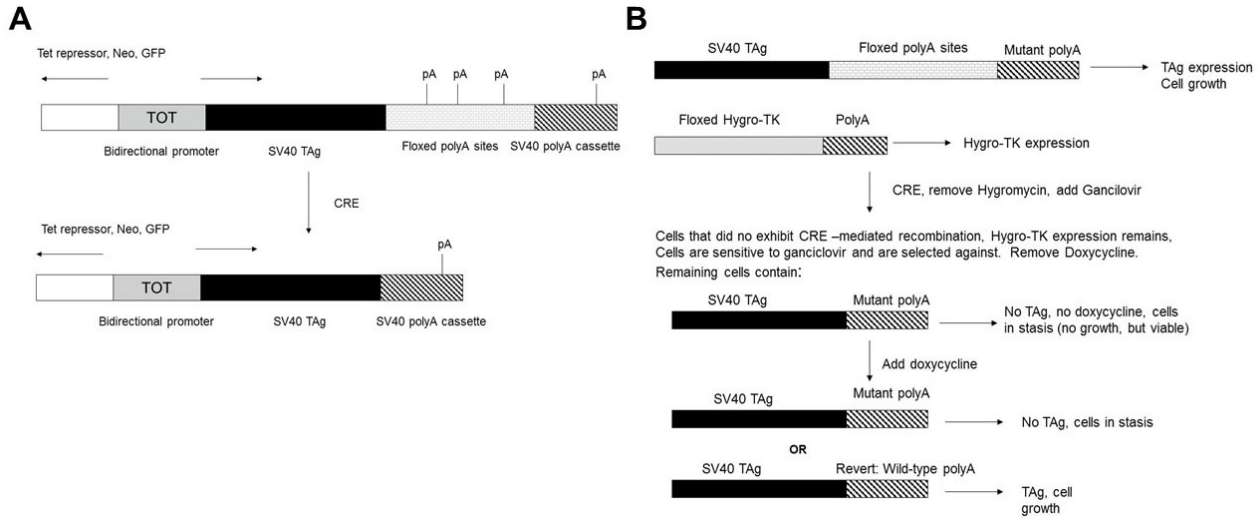


Figure 2. Preliminary data on system characteristics. (A) Repression of TAg expression after CRE-Lox excision of wild-type polyA cassette. (B) Doxycycline-dependent cell proliferation. Gray squares show induction; black diamonds are controls. (C) Doxycycline-dependent clonal cell growth and reanimation. Induced (F+) and not induced (F–) cells are shown. For samples F3 and F6, cells were incubated without doxycycline for 3 or 6 days, respectively, and then doxycycline was added to the media. (D) Typical cellular phenotypes from samples F+ (with doxycycline) and F– (no doxycycline) are shown.

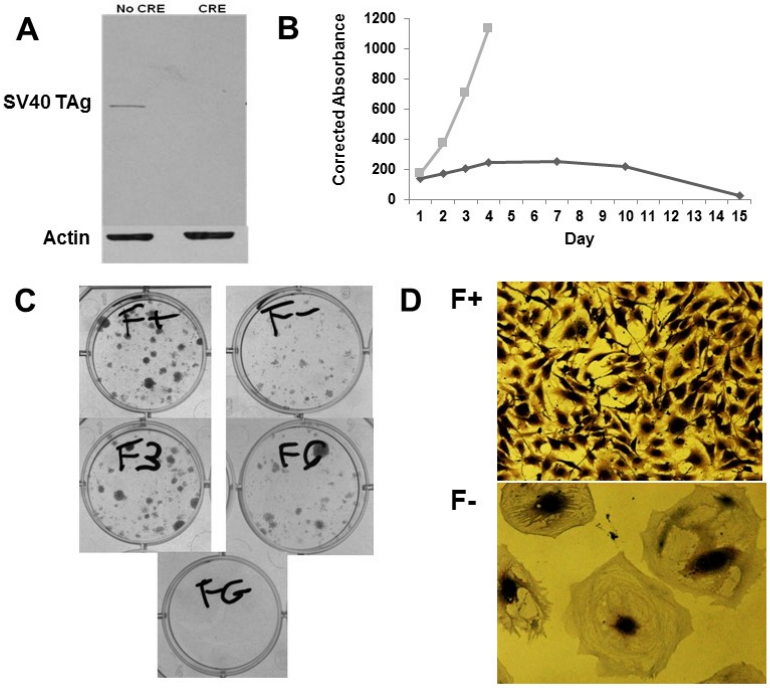


Figure 3. Sequencing data for clone F. Sequencing of F clone target polyA site; PCR product sequencing from genomic DNA. Relevant AACAGG sequence is in larger, bold font, and underlined.

>F5650-SP6_F10.ab1
NNNNNNNTGNTGANNNNGNANNNCNNCAACTAGAATGCAGTGAAAAAAATGCTTTATTTGTGAAAT
TTGTGATGCTATTGNTTTATTTGTAACCATTATAAGCTGC**AACAGG**CAAGTTAACAACAACAAT
TGCATTCATTTTATGTTTCAGGTTCAGGGGGAGGTGTGGGAGGTTTTTTAAAGCAAGTAAAACCTC
TACAAATGTGGTATGGCTGATTATGATCCTGCCTCGCGCGTTTCGGTGAACGGTGAAAACCTCTGA
CATCCCTTTAGTGAGGGTTAATCATGCTCGAGCAGCTCCCGCCTAATTAAGGCAGTCACGTAGCGA
TAGCGGAGTGTACTGCATTAATGAATCGGCCAACGCGCGGGGAGAGGCGGTTTGCGTATTNANNA
AANNAAAAACCNCNNGCNNCTGCTAAAACAACTTCCTGNGNCCGGCCTCCTGGTNGANGNNNNG
NAAAAAATGANAAAAAAAAAAANGNNCCTTTCTNGCGNGNGGNGCNTANNNTACNANATCGNTNT
TNNGATCTATAANAANANNATNNNGCGNNATGNTNGNATAGNCNGCGNGNNGNNNNNANTATNTA
NNTNNNNNNNNGNNNGNGGGGGGGG

Figure 4. Adaptive mutation pilot experiment for cell growth treatment methodology. Lack of growth in small-scale pilot experiment demonstrates that system is working as expected, without significant nonspecific clonal growth.

Polyadenylation Site Mutations

A number of different mechanisms are postulated to lead to an adaptive response affecting gene expression: point mutations (eg, via base tautomerism), frameshift mutations, insertions and deletions, and epigenetic alterations. Although these possibilities can be potentially explained by classical mechanisms, we have a particular interest in evaluating the potential for a quantum biological explanation for adaptive mutation phenomena. The possibility of a quantum mechanism has informed our choice of experimental design, which is based on reversible mutation of polyadenylation (polyA) site signals.

In our experimental system, we would evaluate a polyA site mutation that can be reversed via base tautomerism to wild-type functional sequence. Thus, in the proper environment, a change in the cell state becomes irreversible due to clonal growth; this cell state is associated with a particular DNA sequence that allows for gene expression resulting in clonal growth. For example, consider cell state A_1 characterized by a DNA sequence (ie, the original mutant polyA site) that does not allow for cell growth. In contrast, cell state A_2 is characterized by the countermutation of the polyA site that restores TAg expression and allows for clonal cell growth. In the proper environment (presence of doxycycline), cells in state A_2 undergo clonal growth, which is an irreversible change allowing cell state A_2 to be observed. Because cell state A_2 is associated with the DNA sequence allowing for cell growth, that sequence (of the polyA site) would also be observed when cellular DNA is assayed.

The specific mutations to be evaluated in this study are supported by Cooper's [31-33] analysis of T4 bacteriophage mutation data. Cooper identified G and C bases as particularly sensitive to exist as a lowest-energy state consisting of a linear combination (ie, superposition) of G-C isomers [31-33]. Thus, the polyA site hexanucleotide sequences would incorporate 1, 2, or 3 mutant bases compared to the wild-type consensus sequence (AATAAA); these mutant sequences are AACAAA, AACAAG, or AACAGG. We chose to use base C in position 3 and base G in positions 5 and 6 because (1) mutations in these positions repress 3′ end processing [30] and (2) C to T and G

to A conversions via base tautomerism [31-33] can restore the wild-type AATAAA hexanucleotide sequence. Thus, for a hexanucleotide sequence of AACAAA, the DNA template strand is TTGTTT; with RNA transcription, the mutant polyA site produced is AACAAA. However, if the G of the template strand is in the enol form (base tautomerism), it can base-pair with U resulting in wild-type AAUAAA sequence in the transcript. This would, in turn, restore wild-type 3′ end processing, TAg gene expression, and clonal cell growth. The mutation can also be reversed through DNA replication. Again considering the DNA sequence AACAAA, the complementary DNA strand is TTGTTT; the G in the enol form would base-pair with T restoring AATAAA. The same process can occur with G in positions 5 and 6 of the hexanucleotide sequence (AACAAG or AACAGG); thus, imino C in the complementary strand would base-pair with A, again restoring AATAAA.

Polymerase chain reaction (PCR) analysis, which has been used to probe DNA quantum superposition in vitro [34], also supports utilizing base tautomerism as the underlying rationale for the specific mutations to be used in our study. A study of the effects of primer-template mismatch on PCR efficiency determined that tautomeric G-T and C-A mismatches minimally affect PCR amplification; other types of base pair mismatch diminish amplification to a far greater extent [35]. Although that PCR study did not focus on molecular mechanisms, the findings are consistent with quantum-mediated base substitution being amplified by the PCR amplification process. Successful PCR amplification may be a “potential well” that “captures” base tautomerism (enol G and imino C) resulting from the linear combination of G-C isomers [31-33].

With respect to our pilot experiments, the triple mutation AACAGG exhibits the greatest inhibition of 3′ end processing and the greatest reduction in gene expression compared to wild-type AATAAA [30] (Figure 2). Therefore, we have generated MEF cell clones containing a version of pRITAGKHexa with a triple-mutant polyA site AACAGG (see preliminary data).

Main Experimental Protocols: Fluctuation and Reconstruction Tests

The original Luria-Delbruck bacterial fluctuation test [36] was modified for use in mammalian cells [37-40] and can be used to distinguish between random and adaptive mutations. First, multiple samples from a single large culture (SLC) of the stably transfected MEF cells would be infected with Ad-CRE virus, plated out, and tested for colony growth as described previously. Then, separate multiple replicates (SMR) of the MEF cells would be produced; each replicate would be infected with Ad-CRE, plated out, and evaluated for colony formation. Cells would then be treated with doxycycline; as controls, we would culture 1 set of cells without doxycycline and this control should exhibit no clonal growth.

For both the SLC and SMR samples, numbers of resulting colonies from each set of replicates would be evaluated for the variance vs the mean. For the SLC samples, because the multiple samples come from the same single, original MEF cell culture, the variance would be similar to the mean with respect to number of colonies observed. However, for the SMR samples, each replicate is derived from an independent expansion of the MEF cell clone. Therefore, the relationship between the variance and the mean of colonies from these multiple independent replicates would depend on the type of mutation. For the SMR samples, a large variance vs mean would suggest random (and preexisting) mutation. The explanation for this finding would be that random mutations occur in the replicates at different times during cell culture expansion, so that each replicate would have a markedly different number of mutation-containing cells; hence, this would result in a large variance in colony formation. In contrast, a variance approximately equal/similar to the mean would suggest adaptive mutation. The explanation for this finding is that each replicate culture starts the experiment in the same condition of having approximately zero cells with mutation and the selective pressure induces the adaptive mutation at the same rate in each replicate culture. Thus, the number of colonies formed would be similar between cultures, resulting in a variance similar to the mean [36].

Reconstruction tests (reviewed in [17]) would be performed to determine whether late-forming cell colonies are (1) the result of (random or adaptive) mutations that occurred during the selection process or (2) are slow-growing cells with random mutations that occurred before Ad-CRE infection and before doxycycline exposure. Thus, cells from late-appearing colonies would be isolated and replated, and the clonal growth experiment would be repeated. As controls, cells from early-appearing colonies would be similarly replated. We would determine the time required for cells to form visible colonies of the same size as those from the original experiment. If cells from the early-appearing and late-appearing colonies grow at approximately the same rate in the reconstruction test, this means that late-appearing colonies were due to late-appearing (ie, during selection) mutations. However, if cells from later-appearing colonies exhibit slower growth kinetics than cells from early-appearing colonies, this would suggest that these are colonies derived from cells with mutations preexisting the selective pressure and that these cells have an innate slower growth rate.

In addition to clonal growth assays, cell proliferation can also be measured with the Quick Cell Proliferation assay kit, which we have already used to measure doxycycline-dependent growth of stably transfected MEF cells (Figure 2). It allows for high-throughput analysis of cell growth at defined time points in a 96-well format and the reagent used in this kit has limited toxicity and does not stain cells. Therefore, subsequent to measurements of cell growth, cells can be available for isolation of nucleic acids/proteins.

Evaluation of Possible Mutator Phenotypes and DNA/RNA/Protein Analyses

If adaptive mutation is observed in the aforementioned experiments, mechanism of action would be analyzed by (1) for undirected adaptive mutation, we would evaluate whether changes in the expression of error-prone vs high-fidelity DNA polymerases, as well as MMR proteins, influences the general mutation rate in cells under selective pressure and (2) for directed adaptive mutation, base tautomerism resulting in base substitution mutation would be analyzed by DNA sequencing.

Evaluation of nonspecific adaptive mutations (eg, the "mutator phenotype") would be as follows. First, reversible immortalization of MEF cells has not been shown to lead to random mutations allowing for growth because cells in the absence of TAg expression remain quiescent until TAg expression is restored [29]. Therefore, mutations that allow for cell growth in a manner independent of TAg expression have not been observed in previous studies of MEF cell reimmortalization. Second, we would determine polyA site sequence data for the MEF cell clones. If cell growth is observed even in the presence of the original inactivating mutation, this would suggest that an unknown, likely undirected, mutation activated clonal cell growth. However, if all MEF clones exhibit the predicted countermutation of the polyA site, this would support the directed mutation mechanism. Third, we would conduct hypoxanthine phosphoribosyltransferase (HPRT) assays, a commonly used method to determine increased mutation rates. Cells resistant to 6-thioguanine (6-TG) result from a random mutation in the *HPRT* gene, and any increase in 6-TG resistant cell growth is indicative of an increased undirected mutation rate. Cells should not exhibit enhanced resistance to 6-TG if directed mutation restores the wild-type polyA site sequence. However, if countermutation at the target polyA site is the result of an undirected increase in the mutation rate, then enhanced 6-TG resistance likely will be observed.

If undirected adaptive mutation is observed, we would evaluate whether the mechanism of mutation is through upregulation of error-prone DNA polymerases and downregulation of high-fidelity DNA polymerases and MMR proteins as has been reported in a prostate cancer cell line [16]. Protein lysates would be isolated from expanded cell colonies, and Western blot analysis performed to measure levels of (1) error-prone DNA polymerase (Pol ι), (2) high-fidelity DNA polymerases (Pol δ and Pol ε), and (3) MMR proteins (MSH6, PMS1, PMS2, and MLH1), as previously described [16].

If directed adaptive mutation is observed, nucleic acid sequence analyses would contribute to our understanding of the base tautomerism mechanism by which the mutation is generated. DNA and RNA would be isolated from cell colonies and/or from cells analyzed with the Quick Cell Proliferation assay kit. PCR (DNA) and reverse-transcription PCR (RNA) would be performed to amplify the target polyA region, followed by sequencing of the PCR products. Thus, we would determine whether cells exhibit reversal of the polyA site mutation. In addition, protein lysates would be isolated from expanded clonal growth and TAg protein expression would be measured by Western blot analysis. We would also perform PCR to confirm that the CRE-mediated excision took place and that the wild-type polyA cassette was removed from the TAg expression plasmid.

Proposed Expected Results

Our primary expectation is that directed adaptive mutation would be observed and that the predicted base substitution mechanism would be confirmed by sequencing. We predict that after Ad-CRE infection and the addition of doxycycline, the cells will remain quiescent for a period of time and then commence clonal cell growth. It is possible that no adaptive mutation would occur because either (1) no clonal growth will occur or (2) growth will occur, but will be consistent with random mutation. Another possibility is that the undirected mutator phenotype form of adaptive mutation will be observed. If this occurs, we expect to find altered expression of error-prone vs high-fidelity DNA polymerases and MMR proteins [16]. It is possible that the triple mutation construct will present a mutation threshold that is too difficult to overcome via reverse mutation. In the event that no reversion of mutation occurs with the triple mutation, we would repeat the experimental design with the single and double mutation constructs (see discussion of polyA mutations).

Alternative Approaches

One alternative approach to evaluate the possibility of directed adaptive mutation in mammalian cells is to use a frameshift mutation model in which a change in base sequence can confer resistance to a cytotoxic selective agent, thus allowing for cell growth/colony formation. We would use a plasmid that contains a thymidine kinase-neomycin resistance (TK-Neo) fusion gene and a sequence insert in the TK cassette that causes the subsequent Neo sequence to be out of frame for protein translation [41,42]. Frameshift mutation would alter the elongated sequence insert and restore Neo expression, resulting in resistance to the selection agent G418 [41,42]. The plasmid to be used for these experiments also has a constitutively expressed hygromycin selection cassette; thus, the plasmid can be stably transfected into mammalian cells. Cells would be exposed to a killing concentration of G418 and only cells with a frameshift mutation would express TK-Neo and exhibit G418-resistant clonal growth. The experimental design at that point would be similar to that described previously for the MEF cell studies. If the basis-dependent selection hypothesis is correct, then the frequency of mutation would be greater in the presence of the selective agent. Further, if we put another constraint on cell growth (eg, presence of serum in the tissue culture medium), then we would expect greater rates of specific mutation in the targeted TK-Neo sequence in the presence of the environmental condition that allows for cell growth (ie, presence of serum).

Another possible experimental design has been described elsewhere [27] and uses induction of bacteriophage lambda from the lysogenic to lytic form. This is a prokaryotic system and is, therefore, not an alternative approach for (eukaryotic) mammalian cells. Nevertheless, this prokaryotic approach may be useful in identifying directed adaptive mutation in a system well suited for rapid high-throughput analyses. Thus, for example, we can consider mutations that render the prophage unable to switch to the lytic pathway. The first mutation could be a temperature-sensitive mutation in a regulatory gene, such as cI repressor, whereas the second mutation would be in a gene required for the next step in induction (eg, antiterminator N). The first mutation would be controlled as part of the experiment (eg, changing the temperature to influence cI expression), whereas alteration in the second mutation (ie, countermutation restoring wild-type sequence and gene expression) would occur in a manner not directly influenced by the experimenter. According to the hypothesis of basis-dependent selection, reversion of the second mutation to active wild type would occur more frequently when the cI repressor is inactivated by the

temperature-sensitive mutation. In other words, a mutation that allows for lytic induction would occur more frequently in circumstances in which lytic growth is permissible (no cI repressor activity).

Purpose of Paper

Directed adaptive mutations arise when selective pressure induces targeted mutations that specifically influence the adaptive response; identification of directed adaptive mutation would have profound practical significance for a wide variety of biomedical problems, including disease development and resistance to treatment. The possibility of directed adaptive mutation has not been experimentally assessed due to the lack of an appropriate experimental system. We believe that this fundamental methodological deficiency is addressed by our proposed system; thus, this manuscript describes a possible experimental approach to determine whether directed adaptive mutation occurs in mammalian cells. This approach is based on our model of basis-dependent selection describing a quantum biological mechanism of adaptive mutation.

This project is currently inactive due to lack of funding; the data we have are preliminary, derived from a small number of pilot experiments. However, a major objective of early reports is to discuss projects that have, for whatever reason, not produced publishable data, but are worthy of discussion and analysis because of novel experimental designs, innovative protocols, and/or previously unexplored hypotheses. Thus, we outline the project's rationale and experimental design to stimulate discussion and analysis, and lay the foundation for future studies in this field. Readers can identify strengths and weaknesses of our hypotheses, overall approach, experimental design, and specific research protocols. Thus, critical analysis from the scientific community may lead to improved methodologies for identifying adaptive mutation and for evaluating the validity of basis-dependent selection.

Methods

Plasmids

The pRITAGKHexa plasmids were constructed as follows. The plasmid pGKNeotpAlox2 (Addgene) was digested with *Eco*R1 and *Hin*dIII, Klenow treated, and religated to yield pGKDelta, lacking the neomycin gene. pRITAGK was constructed by ligating the *Nhe*I (Klenow-treated)-*Pac*one large fragment of pRITA with the *Nhe*I-*Pac*I small fragment of pRITA and the *Sac*I (Klenow-treated)-*Nhe*I fragment of pGK delta containing the floxed SV40 late polyA sites. pRITAHexa plasmids were constructed by ligating the *Bgl*II-*Pac*I large fragment of pRITAGK the *Nhe*1-*Bgl*II fragment of pRITAGK containing the floxed SV40 late polyA sites and the *Nhe*I-*Pac*I small fragments from constructs obtained from IDT Technologies, containing a cassette of wild-type (AATAAA) SV40 late polyA hexanucleotide sequences or one of the following mutations: AACAAA, AACAAG, or AACAGG. This formed plasmids pRITAGKHexa-WT, pRITAGKHexa-C, pRITAGKHexa-CG, and pRITAGKHexa-CGG, respectively. This cassette contains *Xho*I sites for screening and SP6 and T3 sites for sequencing analysis of the polyA cassette, distinguishing this polyA site from those in the floxed pGK sequences. To construct p3.1hTKΔNeo, the Hygro-TK plasmid from Alex Bortvin was digested with *Not*I to remove the hygromycin-thymidine kinase cassette. This was inserted into pcDNA3.1-, cut with *Not*I, and treated with calf intestinal alkaline phosphatase. To remove neomycin-resistance function, the bulk of the neomycin gene was removed from the resulting construct by digestion with *Bst*BI and *Nar*I, Klenow treatment, and religation.

Stable Transfection: CRE-Lox Excision of Mixed Cell Population

The plasmids pRITAGKHexa-WT, pRITAGKHexa-C, pRITAGKHexa-CG, and pRITAGKHexa-CGG were linearized with *Pac*I and p3.1hTKΔNeo was linearized with *Mfe*I. Each of the pRITA-based plasmids was stably cotransfected by nucleofection into MEF cells along with p3.1hTKΔNeo and selection was performed with 400 µg/mL G418 and 150 µg/mL hygromycin.

MEF cells stably transfected with the triple-mutant (CGG) pRITAGKHexa plasmid and Hygro-TK plasmids were infected or mock infected with Ad-CRE adenovirus at a MOI of 1000. Cells exposed to Ad-CRE were treated with 50 µM ganciclovir to eliminate cells that were not infected and did not undergo CRE-Lox excision. Three days later, protein was isolated and Western blot analysis performed with anti-SV40 TAg and antiactin antibodies.

These cells were then subjected to clonal selection and individual clones were grown and tested as described subsequently for growth characteristics.

Doxycycline-Dependent Cell Proliferation

MEF cells were plated out at 4000 cells/well in a 96-well plate with 4 µg/mL doxycycline, 400 µg/mL G418, and 100 µg/mL hygromycin or hygromycin alone. Cells were assayed with the QuickCell Proliferation kit (Biovision). For doxycycline-dependent clonal cell growth and reanimation, 500 MEF clone F cells were plated in a 6-well plate in the presence of 4 µg/mL doxycycline, 400 µg/mL G418, and 100 µg/mL hygromycin or hygromycin alone. All cells were grown with a 1:1000 dilution of Fungizone. At the end of the experiment, colonies were stained with crystal violet.

Polymerase Chain Reaction and DNA Sequencing

PCR was performed with a Promega PCR core kit, with a hot start with the following parameters: 1 cycle of 95 °C for 6 minutes; 50 cycles of 95, 56, and 72 °C for 30 seconds each; and 1 cycle of 72 °C for 10 minutes. The following primers were used: X6 (CTCGAGAGGATTTAGGTGACACT) and RGKup (CAATACGCAAACCGCCTCTC). Sequencing of PCR products was performed by Genewiz with the P3 alternative sequencing protocol (custom unprocessed PCR sequencing).

Adaptive Mutation Pilot Experiment

A total of 150,000 MEF clone F cells were plated in a 6-well plate in the presence of 4 mg/mL doxycycline, 400 µg/mL G418, and 100 µg/mL hygromycin. The next day, cells in media without the aforementioned added reagents were infected with Ad-CMV-CRE virus at a MOI of 1000. After 24-hr infection, cells were trypsinized and replated onto a 15-cm dish with 400 µg/mL G418 and 50 µM ganciclovir. After a further 24 hr, 4

μg/mL doxycycline was added to the media. Cells were incubated with doxycycline, G418, and ganciclovir (with regular changing of media) until the end of the experiment; at the end of the experiment (19 days postreplating), colonies were stained with crystal violet. All cells were grown with a 1:1000 dilution of Fungizone except during the 24-hr virus infection.

Results

Preliminary Data Characterizing the Experimental Design

Plasmids, Stable Transfection, and CRE-Lox Excision of Mixed Cell Population

MEF cells were stably transfected with the pRITAGKHexa plasmid (Figure 1) containing the downstream polyA site with the triple-mutant AACAGG hexanucleotide sequence, and cotransfected with the floxed Hygro-TK plasmid. After G418 and hygromycin selection, we performed a pilot experiment in which CRE was delivered by adenovirus (Ad-CRE), resulting in excision of the floxed sequences in the stably transfected plasmids. Cells were then treated with 50 μM ganciclovir, which killed those cells that did not undergo CRE-mediated excision of the floxed genes. The remaining living cells were those that were successfully infected with the Ad-CRE virus. Western blot analysis confirmed downregulated TAg expression because of inefficient 3′ end processing of the remaining mutant polyA site (Figure 2).

Doxycycline-Dependent Cell Proliferation

We then performed clonal selection to identify a stably transfected MEF cell clone that exhibits doxycycline-inducible expression with rapid cell growth in the presence of doxycycline and minimal growth in the absence of doxycycline. Clone F exhibited proliferation in the presence of doxycycline (Figure 2, gray squares) and exhibited minimal nonspecific proliferation in the absence of doxycycline (Figure 2, black diamonds).

Although proliferation assays are a reasonable screening approach to find an appropriate clone, the main metric for this study is clonal growth. Therefore, we performed clonal growth assays on clone F (Figure 2). In the absence of doxycycline (F–), we observed scattered individual cells or larger clusters of cells mostly exhibiting varied forms of senescent phenotypes, including very large flattened cells or elongated cells. These phenotypes are characteristic of senescent fibroblast cells. In the presence of doxycycline (F+), we observed fast-growing large and dense colonies of phenotypically normal fibroblasts; these types of colonies are the endpoint for a positive signal of clonal cell outgrowth. After 3 days cultured without doxycycline (F3), clone F cells could be reanimated to form colonies of phenotypically normal cells, similar to what is observed with F+. After 6 days cultured in the absence of doxycycline (F6), cells began to lose their ability to be efficiently reanimated; however, after exposure to doxycycline, a few colonies of dividing normal cells were observed. F3 and F6 samples exhibited a greater proportion of senescent cells and fewer dividing colonies compared to F+ as a consequence of the periods cultured in the absence of doxycycline (data not shown). Clone F cells are, as expected, sensitive to ganciclovir; thus, when cultured in the presence of both doxycycline and ganciclovir (FG), no colonies were observed (Figure 2).

Polymerase Chain Reaction and DNA Sequencing

Clone F was further characterized by sequencing the target polyA sequence from PCR-amplified genomic DNA, similar to what we planned for our experimental protocol. The expected mutant hexanucleotide sequence was observed (Figure 3).

Adaptive Mutation Pilot Experiment

We subsequently conducted a pilot experiment of the basic adaptive mutation experimental protocol outlined in Figure 1. Given the number of cells used in this experiment (150,000 cells originally plated and allowed to grow overnight), no colonies were observed after approximately 3 weeks (Figure 4). This experiment was repeated twice with similar results (data not shown). These findings support the stringency of our experimental system. Thus, if the system were “leaky” (with cells that escaped both infection/CRE recombination and ganciclovir treatment), then cell colonies would likely have been observed given the number of cells plated. Further, these findings are also consistent with a very low frequency of putative adaptive mutation events because no clonal outgrowth was observed with 3 replicates of this pilot experiment. Therefore, to assay for adaptive mutation, a greater number of cells (eg, millions) will be required for assay in each replicate of the experiment.

Summary

In general, clone F exhibits a number of properties suitable for our experimental design; however, the characterization of even more optimal clones likely would be required. For example, clone F exhibits some degree of doxycycline-independent growth after plating (Figure 2), similar to several other clones tested (data not shown); in contrast, a fully optimized MEF cell clone would exhibit absolutely no cell growth in the absence of doxycycline.

Discussion

Summary

This paper is an early report of a proposed project aimed at evaluating the hypothesis of basis-dependent selection as a mechanism generating directed adaptive mutation in a mammalian cell model system. We have outlined the experimental design, expected results, and have presented preliminary data from pilot experiments characterizing the fundamentals of the proposed system. Thus, readers can identify strengths and weaknesses of our experimental design, leading to improved methodologies for identifying adaptive mutation and for evaluating the validity of basis-dependent selection.

Relevance of Model Systems

One potential criticism of our experimental approach is that model systems are artificial and do not reflect the full complexity of endogenous genes and endogenous microenvironments. However, we believe that a model system approach is the most efficient initial screening method for identifying a heretofore-unknown biological mechanism. The

complexity of endogenous systems introduces many variables that would complicate the evaluation and optimization of our methodology, and would also complicate the definitive identification of directed adaptive mutation. By reducing variables, model systems more effectively allow for the identification of novel biological processes, particularly those suspected to occur at a low frequency. In vitro models are often used to characterize complex biological systems and generally have significant predictive value when findings are subsequently applied in vivo [43]. Thus, the use of model systems for the initial characterization of directed adaptive mutation is warranted.

Further Possible Future Studies

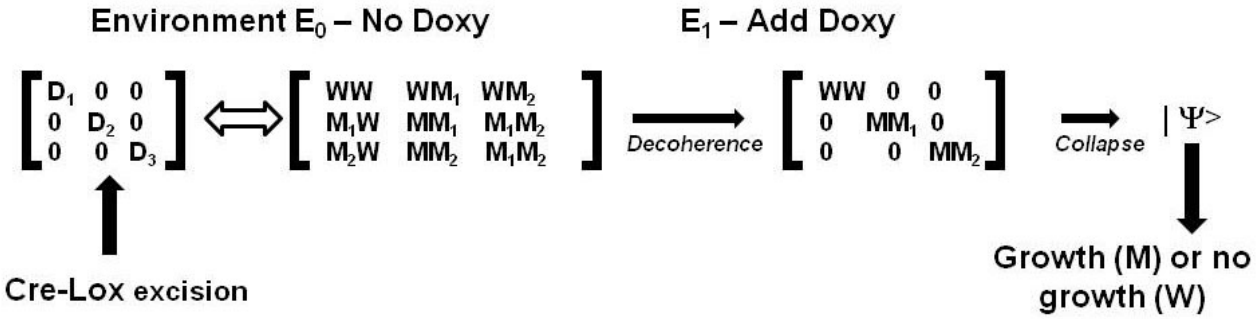
If our proposed project produced findings consistent with undirected adaptive mutation, we would subsequently evaluate therapeutic approaches to reduce the increased mutation rate. For example, siRNA-mediated knockdown of error-prone DNA polymerases, and/or exogenous overexpression of high-fidelity DNA polymerases and MMR proteins could restore a wild-type cell phenotype. If the findings were consistent instead with directed adaptive mutation, we would then determine the general applicability of the phenomenon by evaluating other experimental systems. For example, we would investigate whether directed adaptive mutation contributes to cancer initiation/progression or whether cancer cells use directed adaptive mutation to evade anticancer therapy. Evaluating adaptive mutation during development of a multicellular organism (eg, earliest stages of Drosophila development) is another potential future approach. We would also assess whether targeted alteration of the cellular microenvironment can induce specific, therapeutically favorable mutations (eg, those that increase sensitivity to treatment or which enhance terminal differentiation of neoplastic cells).

Significance

Adaptive mutation has significance for a wide variety of biomedical problems, including carcinogenesis, development of resistance to treatment for cancer and other diseases, and modulation of cell/tissue plasticity in experimental and/or clinical approaches for gene therapy, stem cell therapies, or tissue regeneration. Directed adaptive mutation could force a reevaluation of certain drug-based therapeutic strategies. For example, our model of adaptive mutation [27] requires that cells have sufficient time to "explore" the space of possibilities. Thus, to prevent directed adaptive mutation, it would be important to use cytotoxic methods to kill cells quickly [27]. Conversely, longer-duration treatments that allow cells to explore the adaptive possibilities could be used to promote clinically favorable mutations (eg, those causing differentiation and/or apoptosis). There is also practical importance in distinguishing directed vs undirected adaptive mutation [27]. Understanding directed adaptive mutation may lead to approaches that use targeted manipulation of the microenvironment to prevent or induce specific mutations and consequent changes in gene expression. In contrast, therapeutic approaches for undirected adaptive mutation would be limited to suppressing mutation rates.

Findings from our proposed experimental system may also have implications for the theory of basis-dependent selection previously described [27]. Thus, for the sake of theoretical evaluation, a quantum mechanical density matrix formalism describing the experimental design and potential results is shown in Figure 5.

Figure 5. Quantum representation of the experimental design resulting in adaptive mutation. The "D" states represent preferred cell states in an environment lacking doxycycline and after CRE-Lox excision. The "W" states represent the cell state that is wild type from the perspective of the original inactive polyA site, whereas the "M" states represent cell states in which mutation (eg, the polyA site) allows for cell growth in the presence of doxycycline.



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Authors' Contributions

MB conceived the experimental design, performed the majority of the experimental work described, and wrote the manuscript. CRC assisted in the isolation of the stably transfected MEF clones used to generate the preliminary data and also performed Western blot analysis on SV40 TAg expression. TM provided conceptual assistance for the conditional reimmortalization approach used in the experimental design.

Conflicts of Interest

TM has filed for a patent covering the conditional immortalization technology. The other authors declare no conflicts of interest.

Multimedia Appendix 1

Powerpoint presentation of key concepts of basis-dependent selection.

[PPT File (Microsoft PowerPoint Presentation), 562KB - resprot_v3i4e74_app1.ppt]

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Abbreviations

6-TG: 6-thioguanine
GFP: green fluorescent protein
HPRT: hypoxanthine phosphoribosyltransferase
MEF: mouse embryo fibroblast
MOI: multiplicity of infection
PCR: polymerase chain reaction
SLC: single large culture
SMR: separate multiple replicates

SV40: simian virus 40
TAg: large T antigen

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Original Paper

The Evolution of a Professional Practice Forum: Balancing Peer-to-Peer Learning With Course Objectives

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Abstract

Background: The Opioid Treatment Accreditation Course (OTAC) is a mandatory accreditation requirement in New South Wales, Australia, and aims to prepare medical practitioners for the provision of safe and effective Opioid Substitution Treatment to people with opioid dependence. The course has a strong focus on safe prescribing practices and the course design includes a Professional Practice Forum that is engaging for participants and effective at imparting complex ideas and concepts that do not place additional time constraints on already time-poor health professionals.

Objective: The study aimed to use participatory action research methods to develop and evaluate an online Professional Practice Forum that is a key component of the OTAC teaching and learning experience.

Methods: Three evaluation cycles were implemented with three cohorts of participants (N=40) to inform the design and review of the updated OTAC course. Overall, the study relied on participatory action research methods to enhance a sense of online community and to revise the Professional Practice Forum component of the course. Findings from survey feedback and an examination of Web metrics were used to monitor participant learning and were subsequently subject to thematic analysis in order to identify key themes.

Results: The use of participatory action techniques in the redesign of the OTAC course was a successful means of engaging with participants and resulted in four revisions based on feedback from facilitators and participants. The Professional Practice Forum was rated highly and received positive feedback from both moderators and participants.

Conclusions: The use of interactive forums in online learning in an educational module for adult learners can prove extremely valuable as a means for participants to share their expertise and improve their learning outcomes. In particular, the use of sticky and welcome threads were significant features that enhanced interactions between participants and facilitators and resulted in increased quantity and quality of postings. These findings can help inform future researchers on how to develop peer engagement modules that are amenable to assessment and that build an online sense of community.

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KEYWORDS

medical education; peer-to-peer; online learning; formative assessment

Introduction

Overview

Over the last decade, there has been increasing demand for continuing medical education programs that enhance the clinical skills and knowledge of medical specialists and general practitioners [1,2]. This is particularly the case in relation to providing safe treatment for people with opioid dependence because of the widespread stigma associated with opioid use in Australia. At the same time, medical practitioners have high-volume work environments and are often time-poor. Hence, educators need to develop high-quality learning environments and to identify and evaluate learning outcomes to ensure they have utility for participants and can be readily applied into their own practice [3,4]. The current Opioid Treatment Accreditation Course (OTAC) is designed around the core principles of both adult learning and situated learning [5-7]. This approach recognizes the knowledge and experience that participants bring to learning environments, and the literature confirms that this approach contributes to the creation of authentic and "real life" activities and contexts to promote learning [8].

The use of online forums and peer-to-peer learning approaches are now commonplace in e-learning because of their capacity to engage participants and to promote interactive learning environments [9,10]. In addition, the literature confirms that peer-to-peer learning approaches are particularly appealing in online medical education because they emphasize individual autonomy and participants are able to take responsibility for their own learning experiences [11]. Furthermore, collaborative educational environments allow adult learners to identify their existing knowledge and determine their future educational needs [12].

Nevertheless, the literature identifies high rates of attrition from online courses and that educators need to ensure the online environment is engaging and promotes a sense of community between participants [13]. Although challenging, a systematic review of 57 studies on e-learning for health professionals and students demonstrated that well-designed e-learning packages are learner centric and share responsibility between trainers and learners [14]. A significant challenge to building a thriving online community is the time and effort required to build a sense of community that requires enabling efforts more than moderation of participant activity [15]. This paper describes the development and review of an online course and Professional Practice Forum for clinicians who wish to be accredited to dispense Opioid Substitution Treatment in New South Wales (NSW), Australia.

Background to the Opioid Treatment Accreditation Course

The treatment of opioid dependence is often challenging for medical practitioners and prescribing opioids in NSW occurs within a regulatory framework that requires clinicians to have approval from the NSW Health Department to prescribe to each patient [16]. The OTAC can be completed as either an online course or as a 1-day face-to-face workshop. In 2009, addiction medicine was formally recognized by the Australian government as a medical specialty. Given the complex mental and physical health needs experienced by people recovering from opioid dependence, it is anticipated that this recognition will improve the safety and standards of health care for this cohort. It is noteworthy, however, that not all practitioners who complete the OTAC course go on to be active prescribers.

The OTAC course primarily targets general practitioners, who are already heavily targeted for continuing medical education, much of which is online. Hence, medical practitioners have high expectations in relation to the quality of online materials and are well placed as informants for the design and application of online learning programs [2]. In 2011, the Workforce Education and Development Group at the University of Sydney undertook a content and design review of the OTAC and subsequently piloted the program with medical practitioners (N=14) from diverse locations, including regional and rural centers. The aim of the review was to realign the course with the latest approaches in online learning in the medical sector and to ensure that the program was consistent with the principles of adult education that include self-directed learning and knowledge acquisition [14,15]. Subsequently, the OTAC was implemented and evaluated with two more cohorts to further develop and refine the course components and, in particular, the Professional Practice Forum.

Providing participants with opportunities to consult experts and collaborate with their peers was seen as instrumental in exploring their attitudes to prescribing and in developing support and knowledge networks that could be sustained subsequent to their participation in the course. Hence, the updated OTAC consisted of three modules that allowed participants to be self-directed in attaining their learning objectives over the 4-week course duration and a Professional Practice Forum (the third module) that was a mandatory component for satisfactory course completion. This forum was designed to enable participants to be assessed by experienced prescribers (referred to as facilitators) to ensure they had achieved key learning outcomes and to allow participants an opportunity to reflect on and demonstrate the knowledge they had acquired.

The forum provided a platform for participants to collaborate and to reflect on their learning in an online community environment. Two experienced facilitators were recruited to moderate the forum and to formatively assess the extent to which participants had developed their knowledge and skills in line with the core learning principles of the wider course. Given the challenges involved in recruiting busy practitioners for lengthy periods of time, participants were only required to interact with peers and moderators for a maximum of 4 hours. The forum was a largely asynchronous activity, and this allowed participants to access the discussion at a time of their convenience. The forum was made available for a 4-week period and participants had a 5-day period in which they could interact with, and be assessed by, the moderators.

This paper reports on the modification and evaluation of the Professional Practice Forum designed to increase the knowledge and confidence of medical practitioners to become accredited prescribers in the OTAC program. In addition to reviewing and redesigning the course, the study used participatory action

research (PAR) approaches in order to identify the salient factors for sustaining an online collaborative network in the field of opioid substitution. The study hypothesized that the use of PAR to design the Professional Practice Forum would enhance the learning experience and retention of participants in the course. PAR has been described as a collective and self-reflective inquiry that researchers and participants undertake in order to understand and improve on the practices in which they participate and the situations in which they find themselves [17]. In the current study, PAR methods were used to inform the development and refinement of the Professional Practice Forum.

Methods

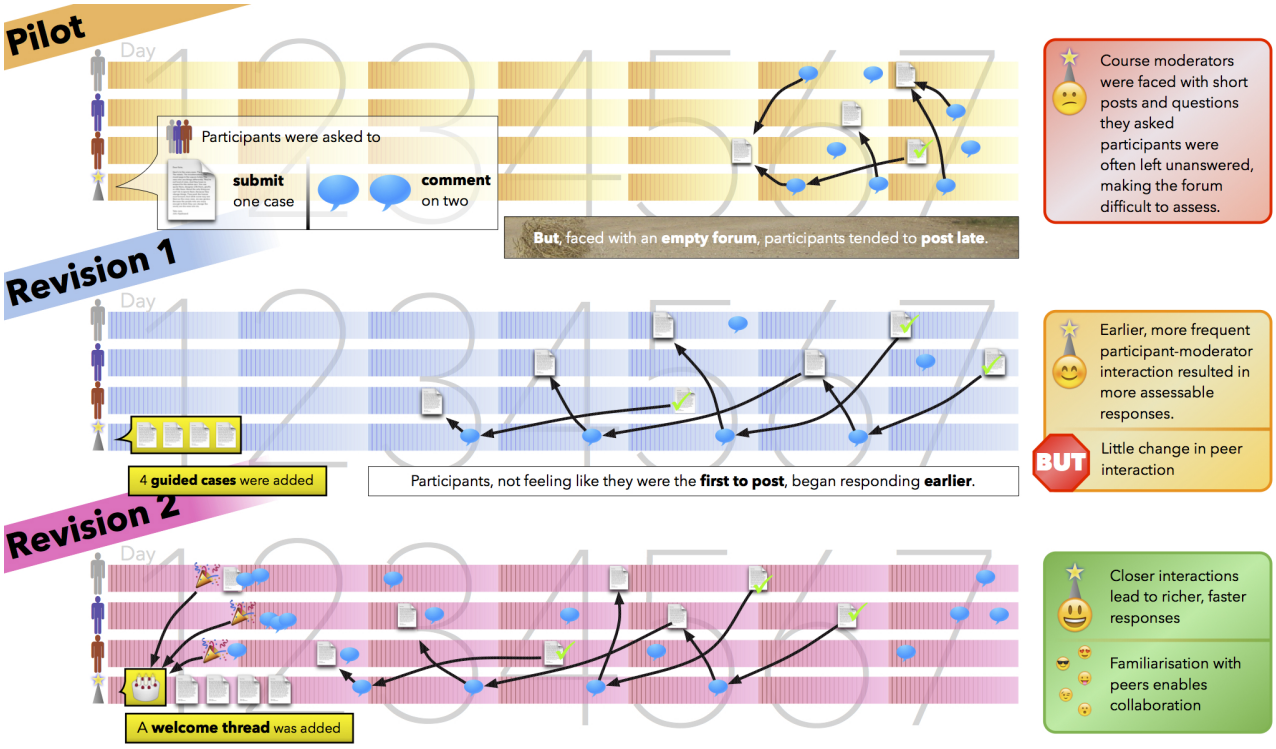
The current study involved three evaluation cycles of the OTAC with three cohorts of participants (N=40) and two facilitators who were also accredited and experienced prescribers. Participants were recruited primarily via email mail-out of course flyers, but some promotion was done via phone. Process and impact evaluations were undertaken after each of the three course iterations. In particular, observational techniques were used to assess processes such as peer interaction and the use of online resources. Analysis of website metrics was also undertaken, including the time that elapsed between the commencement of the forum module and when participants posted their first response. Interactions on the Professional Practice Forum were observed and recorded by the course coordinator in order to review the extent of interaction between participants and between participants and facilitators. Outcome measures were assessed using self-report surveys that asked participants to rate their learning experiences and to comment on their online sense of community. Permission to conduct this study was received from the University of Sydney's Human Research and Ethics Committee.

The Professional Practice Forum was piloted during 2011. At the completion of the first two modules, all participants were emailed by the course coordinator and invited to submit a short scenario to the forum that demonstrated their understanding and application of the previous course modules. In addition, participants were asked to comment on the scenarios of at least 2 peers in order to foster engagement and team problem solving. Subsequent to their completion of the OTAC, participants were asked to respond to an evaluation on their experiences with the forum via an online survey. The survey measured their interest in the Professional Practice Forum format as well as the ease of use and any changes they would recommend. In addition, the forum facilitators were asked to provide feedback on the challenges and benefits of the forum in relation to its utility and how effective it was for enhancing participant knowledge and confidence.

The findings from the pilot study informed revisions to the Professional Practice Forum and the revised OTAC was subsequently implemented with (N=13) participants. This iteration of the course included four "sticky" threads that linked to exemplars of case scenarios, and participants were invited to respond to at least one scenario prior to submitting their own case studies. All other evaluation methods were consistent with the pilot study. A final iteration of the OTAC course was developed and tested in mid-2011 (N=13) This version of the course included a welcome thread that provided participants with an opportunity to access information on the backgrounds and expertise of course facilitators and to post their own biographical information. In addition, a feedback thread was included to enable an accessible and ongoing evaluation tool for future OTAC participants.

Overall, the study used PAR methods to identify strategies for creating a sense of community and to revise the Professional Practice Forum. Feedback from each of the three course iterations was subject to thematic analysis that was undertaken until saturation was reached and clear themes emerged. See Figure 1.

Figure 1. Visualization of the three evaluation phases, which resulted in revisions to improve the educational impact of the Professional Practice Forum.



Results

A total of 40 participants completed all aspects of the OTAC and provided process and impact feedback. Course participants included 14 general practitioners, 6 registrars, 1 psychiatrist, and 2 chief medical officers; 15 participants indicated they were from rural and regional NSW and the remainder were based in urban centers. Findings that emerged from the feedback (surveys and Web metrics) demonstrate that the use of participatory action techniques in the re-design of the OTAC program was a successful means of engaging with participants and resulted in four revisions based on feedback from both facilitators and participants.

In the pilot OTAC, the open-interfaced design of the Professional Practice Forum appeared to discourage participants from making initial posts, which meant the course coordinator had to repeatedly email instructions and reminders to participants. The inclusion of four "sticky" threads in the second iteration of the course reduced the time participants took between commencing the course and posting their first discussion thread. It also allowed more discussion time for moderators and participants and a longer timeframe for peer interaction. In addition, the guidance provided by the sticky cases resulted in more pertinent responses and content between participants as well as lengthier posts that contained more detailed information about treatment options and management.

Feedback from facilitators' observations of the Professional Practice forum indicated that some participants remained hesitant about fully engaging with their peers. This resulted in a third iteration of the OTAC that included a welcome thread. Participants reported that this had a notable impact on their engagement and sense of online community, which was demonstrated by their increased number of posts as well as the frequency and depth of their interactions with each other. The welcome thread allowed participants to introduce themselves and build rapport with their peers. As one participant stated, "I enjoyed this component and it was interesting to see the various challenges that all clinicians have at their point of treatment or engagement".

Overall, participants reported that they enjoyed the OTAC course and that it resulted in improved knowledge about prescribing and managing people with opioid dependence. In particular, they enjoyed the opportunity of engaging with facilitators who had extensive experience in the field. As one participant stated, "It is an excellent teaching session, I learned a lot from seniors". Nevertheless, facilitators did report that their engagement with and observations of the Professional Practice Forum were time consuming and that some participants required considerable encouragement to complete all forum activities. The use of PAR methods resulted in three changes to the OTAC and provided important information on the factors that enabled participation in the forum.

Discussion

Principal Findings

The re-design of the OTAC Professional Practice Forum was informed by literature on autonomous learning [1,2] and collaborative peer learning [11,14]. Most participants were able to commit approximately 4 hours of their time over the 7-day period to engage with forum activities. Nevertheless, facilitators and the course coordinator reported that they had to be proactive to ensure that all participants engaged fully with the course. This highlights the challenges involved in creating an evolving online community rather than choreographing participation. It also highlights the important role of "enabling" rather than

moderating online facilitation. The use of current prescribers as facilitators was an important strategy for engaging participants and for enabling them to actively join the forum.

At the same time, facilitators reported challenges in facilitating the forum, particularly in relation to the administration burden of following up with participants in the first two iterations of the OTAC. Previously, participants had a tendency to post only what was required and to post directly to facilitators, without drawing on the expertise of their peers. The fact that participants increased their engagement with each other in the final OTAC demonstrates that when they are able to familiarize themselves with each other and when they are provided with exemplars of the work that is required, they are much more likely to actively contribute to the forum.

Another challenge with the creation of online forums is ensuring that participants post enough detail in their responses to make them amenable to both formative and summative assessment processes. The incorporation of the four sticky cases into the Professional Practice Forum after the initial pilot was an effective tool for encouraging participants to post comments on the forum with sufficiently detailed responses. This allowed facilitators to more accurately assess participant knowledge and meant there was more time for them to interact with participants and to request additional information. In the first iteration of the OTAC, participants were disinclined to post to the forum and this challenge is consistent with the literature that identifies participant engagement as a significant challenge in online learning [9].

In subsequent revisions of the OTAC that used guided and exemplar cases, participants were observed to make earlier postings and provided more detailed answers, which were noticeably longer and contained more detailed information on patient history and other diagnostic criteria. This demonstrated their depth of knowledge in regard to symptoms, treatment planning, and management. In the final revision of the Professional Practice Forum, the inclusion of a welcome thread had a noticeable impact on peer interaction. The use of the welcome thread provided participants with immediate access to the Professional Practice Forum and enhanced the growth of an online community. This highlights the importance of encouraging early online interaction between participants and recognizing that learning is also a social process. This is consistent with learner- and community-centered approaches to teaching and learning that emphasize the importance of building on participant knowledge, providing and receiving feedback, and self-evaluation.

Limitations

Several limitations to the current study must be acknowledged. The use of purposive sampling and the diverse discipline backgrounds of participants means that the findings are not generalizable and further dissemination and evaluation of the OTAC course is warranted. In particular, future evaluations of the course should include follow-up information on whether participants go on to become active prescribers. At the same time, the sample was sufficient for the development and piloting of the Professional Practice Forum. The reliance on qualitative feedback from participants is another limitation, and the use of validated measures for online sense of community would be beneficial for quantifying the sense of connection between and within participants and facilitators. Furthermore, future evaluations of OTAC should include long-term follow-up with participants to ascertain if changes in their knowledge are sustained.

The use of PAR methods did, however, provide rich feedback on modifications that were required of the OTAC and resulted in an increased number and quality of interactions in the Professional Practice Forum. Given that the study aimed to build and sustain an online learning community and to enhance retention of busy clinicians, the methods used were adequate for the pilot implementation and refinement of the OTAC.

Conclusions

The challenges of developing and sustaining a sense of community in online learning environments are well documented in the literature. However the literature has a particular focus on strategies for enhancing the sense of connection between participants and facilitators. There is still limited research on how to generate peer-to-peer interactions and harness them as a vehicle for developing and implementing online forums. The findings from this study contribute to better understanding of the factors that encourage peer-to-peer learning.

Peer-to-peer engagement in an educational module for adult learners can prove extremely valuable as a means for participants to share their expertise and improve their learning outcomes. The use of a forum module allowed the course designers to use both formative and summative assessment and evaluation processes. Given the relatively small number of participants, however, it is important that the OTAC is tested with a larger cohort of participants to further explore its utility across different geographical contexts where the use of online education may be less familiar. The use of participatory action approaches in the conduct of the current study was highly effective for allowing participants to engage actively in the construction of the Professional Practice Forum.

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Authors' Contributions

Anna Janssen made a substantial contribution to program development, data collection and analysis, and manuscript preparation. Tracy Robinson made a substantial contribution to manuscript preparation. Tim Shaw made a significant contribution to program design and manuscript preparation.

Conflicts of Interest

None declared.

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Abbreviations

NSW: New South Wales
OTAC: Opioid Treatment Accreditation Course
PAR: participatory action research

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Original Paper

Treatment of Infantile Hemangioma in Regional Hospitals With eHealth Support: Evaluation of Feasibility and Acceptance by Parents and Doctors

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Abstract

Background: Since beta blockers became the preferred treatment for infantile hemangiomas (IH), the number of patients eligible for treatment is increasing. Currently treatment of IH with beta blockers is mainly reserved for expert centers, where wait times are lengthening. This demonstrated the need for development of a more efficient and accessible way of providing care for children needing treatment for IH. An eHealth intervention, Hemangioma Treatment Plan (HTP), was developed to treat IH in regional hospitals with online support from an academic doctor.

Objective: Our goal was to evaluate the feasibility of the eHealth intervention by determining its use, acceptance, and usability. By evaluating the feasibility, usage can be predicted and points for improvement can be defined, thereby facilitating implementation of the intervention.

Methods: Parents of children with an IH, presenting between October 2012 and November 2013 at the tertiary expert Center for Congenital Vascular Anomalies Utrecht, requiring treatment with a beta blocker, were asked to participate in the digital HTP. Both parents and regional doctors were sent a study questionnaire. Acceptance and usability of the HTP were evaluated by using the modified Technology Acceptance Model.

Results: A total of 31 parents and 22 regional doctors participated in the eHealth intervention and received the questionnaire, and 25 parents and 15 doctors responded (response rates respectively 81% and 68%). A majority of the parents (96%, 24/25) and the regional doctors (87%, 13/15) considered the eHealth intervention useful in the care for IH. Most parents (76%, 19/25) and over half of the regional doctors (53%, 8/15) found the HTP easy to use. Technical problems using the HTP were reported by 28% (7/25) of the parents and 73% (11/15) of the doctors. The majority of parents (92%, 23/25) felt positive about usage of the HTP during treatment of their child. All regional doctors (100%, 15/15) felt positive about transition of treatment from the tertiary expert center to them, and 93% (14/15) felt positive about using the HTP.

Conclusions: Our eHealth intervention shows good feasibility, especially among parents. Improvement with respect to technical problems, training of regional doctors, and achieving organizational support might be needed for successful implementation in the future.

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KEYWORDS

eHealth; personal health record; e-consulation, tertiary teledermatology; Internet; acceptance, usability; infantile hemangioma; child

Introduction

Infantile hemangiomas (IH) are common benign vascular tumors found in approximately 4-10% of Caucasian infants [1,2]. Most IH have an uncomplicated course, and a general "wait and see" policy is often justified [3]. However, 24% of patients with IH experience complications, like ulceration, bleeding, functional impairment, life-threatening risk, or cosmetic risk, of which 38% need treatment [3]. In 2008, the efficacy of propranolol, a non-selective beta blocker, in the treatment of complicated IH was discovered, and propranolol became the primary treatment of choice [4,5]. Atenolol, a selective beta-blocker, has also been described as effective in the treatment of IH, showing less severe side effects compared to propranolol [6]. Since beta blocker treatment for IH shows less adverse effects and is less invasive compared to previously used treatment options (like systemic corticosteroids, interferon, and vincristine), the number of patients eligible for treatment is increasing [5-7].

Treatment of IH is currently taking place particularly with multidisciplinary expert teams in tertiary academic centers, like the Center for Congenital Vascular Anomalies Utrecht (CAVU), Wilhelmina Children's Hospital, University Medical Center Utrecht, the Netherlands. However, our wait times are lengthening due to the increasing patient flow. This, together with the sometimes long travel distances for parents, shows the need for development of a more efficient and accessible way of providing care for children needing treatment for IH.

eHealth can be defined as the use of all information and communication technology to improve health and health care [8]. In the Netherlands, there is an eHealth intervention to help parents in the diagnostic process of the vascular skin lesion of their child called Aardbeivlek [9]. This eHealth intervention helps parents correctly diagnose and evaluate an IH after completing an eLearning module. To improve health care in children with skin diseases, we have developed the online pediatric Skin House (huidhuis.nl), a digital interactive platform for information, treatment, and exchange of expertise of pediatric skin diseases, which is accessible to patients, their parents, and health care providers. This study describes a part of the pediatric Skin House: a Web-based personalized eHealth intervention called Hemangioma Treatment Plan (HTP) for treating IH. This eHealth intervention consists of a digital interactive platform of information, treatment, and expertise about IH. It also includes a personal health record (PHR) owned by the patient who gives access to the professionals involved. The aim of this eHealth intervention is efficient and easily accessible care for children with IH by making disease knowledge, treatment protocols, and the PHR easily available to both parents and health care providers. By using the eHealth intervention, children with IH can be treated by their medical doctor in a regional hospital with online support of the experts of the CAVU team (tertiary academic care).

The goal of this study was to evaluate the feasibility of this eHealth intervention by determining its use, acceptance, and usability by parents and doctors. By evaluating the feasibility, usage can be predicted [10,11] and points for improvement can be defined, thereby facilitating implementation of the intervention in the future.

Methods

Design

A cross-sectional study was performed to evaluate the feasibility of the eHealth intervention judged by parents and medical doctors.

Participants

Parents

Parents of children with an IH, presenting between October 2012 and November 2013 at the CAVU and requiring treatment with an oral beta blocker, were asked to participate in the digital HTP. Indications for treatment were (risk of) ulceration, (risk of) functional damage, and (risk of) cosmetic damage. Parents who did not have access to a computer were excluded. Other exclusion criteria were no/insufficient knowledge of the Dutch language and complications of the IH requiring specialized multidisciplinary care. Decisions on inclusion and exclusion based on above mentioned criteria were made by an expert member of the CAVU team.

Medical Doctors

Regional medical doctors (pediatricians and dermatologists) were informed about the HTP by a digital mailing and/or by personal invitation. After showing their interest in participation, they were included in our database. Children who needed beta blocker treatment were referred to the regional medical doctor closest to their home/residence. Other inclusion criteria were access to Internet and ability to measure blood pressure (BP) in a young child.

The study was approved by the ethics committee of the University Medical Center Utrecht.

Intervention

Hemangioma Treatment Plan

In order to achieve more efficient and easily accessible care for IH, an eHealth intervention was designed (in Dutch), called HTP. This interactive digital treatment platform consisted of multiple elements providing the following functions: (1) storage and sharing of patient health information (in a digital PHR) through a secured Web-based portal, (2) providing information about the disease and treatment protocols, (3) facilitating communication between parent and the medical doctor (e-consult), and (4) facilitating communication between the regional and academic doctor (tertiary teledermatology).

The purpose of the HTP is first to warrant safe transition of IH treatment from academic doctors to regional doctors by using digital support. Second, its purpose is to involve parents in the care for IH by making information and contact with doctors more efficient and accessible.

The HTP, including a PHR, was developed in collaboration with Patient1, a private company that offers a secured digital health platform. The health platform includes a PHR, professional health care records, digital research center, and information websites like pediatric Skin House. The PHR was compliant with all Dutch rules and regulations with respect to privacy protection and was checked and approved by the Dutch Privacy Protection Authority. Participation in the HTP was free of charge.

Instructions on the use of the HTP were given to the parents verbally and in writing. Parents created an individual account for their child on the PHR website by registering name, birth date, and personal identification number of the child. By using a password, safe uploading of personal information on the website was guaranteed. The PHR account contained information about IH treatment, the HTP, a message-function (for e-consultation and tertiary teledermatology), and a facility to upload photographs and record effect and side effects. Parents created the PHR themselves and gave the medical doctors access to the PHR.

Regional doctors were given instructions on the use of the HTP in writing and sometimes verbally by phone. Regional doctors also registered at Patient1 using name, birth date, and a personal identification code for health care providers in the Netherlands). After verification and authorization by Patient1 and the parents, the regional doctor involved had access to the individual account of the child being treated.

Treatment Protocol

Treatment of the IH was started with atenolol at the tertiary center, after evaluation of possible contra-indications (which included an electrocardiogram) [12]. Follow-up was performed by the regional doctor near the patient's home, using the HTP. At the age of 1 year, all children were seen by the CAVU team of the academic center to decide whether or not to stop treatment. Regional doctors used an IH treatment protocol for follow-up of the child treated for IH, accessible via the HTP. The protocol describes set moments for consultation and instructions on how to monitor effects and side effects of treatment. Prior to each consultation, parents uploaded photographs of the IH of their child, scored the severity of the IH, and completed standardized questionnaires on potential side effects. With the information provided by the parents and findings during the consultation, the regional doctor decided on further treatment policy, guided by the IH treatment protocol. Findings and policy were reported in the PHR of the HTP.

When advice from the academic doctor (expert dermatologist of the CAVU team) was required, the parent or the regional doctor could send a message via the HTP (respectively e-consult or tertiary teledermatology). These questions were answered within 3 working days by the academic doctor. For urgent situations, such as severe side effects of treatment, parents were instructed to contact the academic doctor who was available 24 hours a day by phone. Patients and doctors received automatic notification messages in their personal email inbox when a message was placed in the HTP. Figures 1 and 2 show screenshots of the HTP.

Figure 1. Screenshot of the Hemangioma Treatment Plan (in Dutch) showing the page where parents upload a photo of the hemangioma prior to consultation (fictitious patient in a test environment).

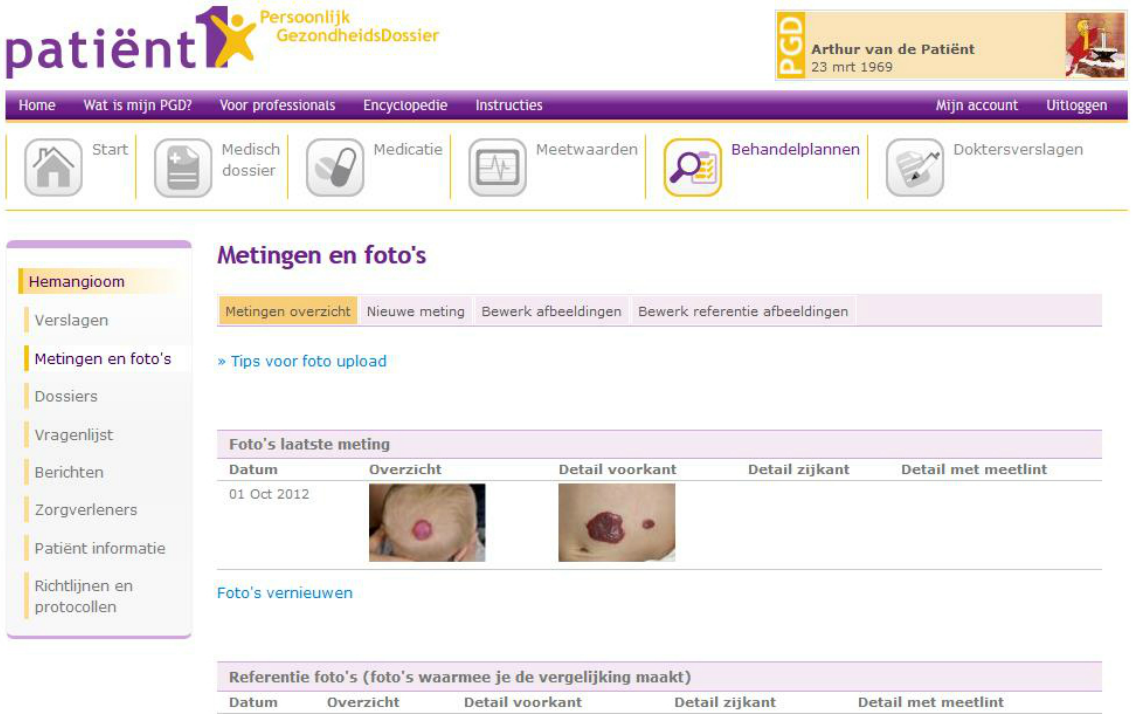
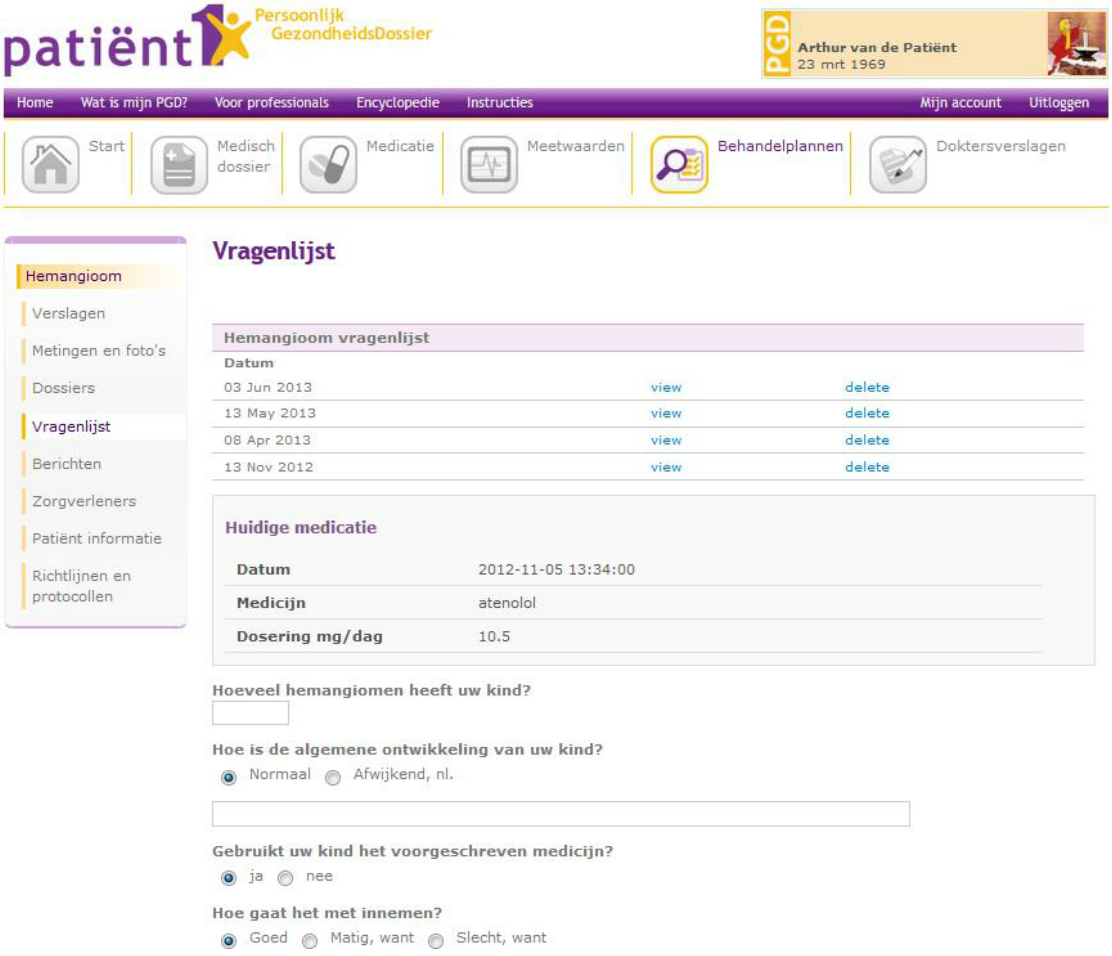


Figure 2. Screenshot of the Hemangioma Treatment Plan (in Dutch) showing the page where parents fill in standardized questionnaires about side-effects prior to consultation (fictitious patient in a test environment).



Variables and Measurement

A questionnaire to evaluate feasibility was developed consisting of the variable use, acceptance, and usability of the eHealth intervention.

The questionnaire was developed based on a modified Technology Acceptance Model (TAM). The TAM model, proposed by Davis in 1989, is based on the Theory of Reasoned Reaction and proposes that Perceived Ease of Use (PEU) and Perceived Usefulness (PU) predict user acceptance of information technology [10,11,13]. Technology acceptance is defined as "an individual's psychological state with regard to his or her voluntary or intended use of a particular technology" [14]. The TAM has been tested for the prediction of adoption of telemedicine by health care professionals and can predict technology acceptance in both obligatory and voluntary usage settings [15,16]. It is suitable for both genders, various age groups, most cultures, and for individuals of all levels of information technology competency [16]. To determine the use, acceptance, and usability of our eHealth intervention, we modified the TAM by adding the dimension "attitude towards use" to the original TAM. Attitude can be defined as "the perception by an individual of the positive or negative consequences related to adopting the technology". Behavioral intention is also determined by attitude, which is influenced by PU and PEU [17]. Questions to evaluate use, acceptance, and usability were developed following the modified TAM.

After at least one consultation with the regional doctor, both parents and doctors were sent a structured study questionnaire. The study questionnaire of the parents and doctors consisted of 38 and 29 questions respectively, grouped into 3 variables (demographic information, use, acceptance and usability) (Table 1). Acceptance and usability were subdivided by the three dimensions of the TAM (PU, PEU, and attitude). On a 3-point scale (agree-no agreement/no disagreement-disagree), we rated 22 questions for the parents and 15 for the doctor. Six and nine questions respectively of the parent and doctor questionnaire could be answered with yes or no, and with the final question, parents and doctors were asked to rate the eHealth intervention on a 0-10 scale (0=very bad, 10=excellent). Apart from the generic questions, all questions contained room to clarify the answer. At the end of the questionnaire, there was an open field for comments and suggestions. Prior to the study, it was determined that an average of 90% of the parents and doctors had to score the items positively regarding feasibility to qualify the eHealth intervention as feasible.

Table 1. Questions used to evaluate feasibility of the eHealth intervention.

Variable	Dimension	Related questions		Example
		Parent questionnaire	Doctor questionnaire	
Demographic information				
		1-6, 9, 11, 13	1-3, 6	Parent: gender, age, education level, residence, treatment indication
				Doctor: gender, age, medical specialism
Use				
		7e-f, 10, 16, 17h	4c-d, 8, 10	Treatment at the regional doctor corresponds with information given by academic center
Acceptance and usability				
	Perceived usefulness	8, 17e, 19, 20a-c	4e, 5, 9f	The e-consult function of the HTP is useful
	Perceived ease of use	15, 17d, 17f-g, 17i-j,	7, 9a-e, 9g-h, 12, 13a-d	The instruction letter of the HTP is understandable and clear
	Attitude	7a-d, 7g, 12, 14, 17a-c, 18, 21	4a-b, 9i, 11, 14	I feel positive about treatment by a regional doctor, with digital support from an expert

Statistics and Analyses

User statistics were recorded. The number of e-consultations, tertiary teledermatology consultations, and responding times were calculated.

Descriptive analyses were used to evaluate the use, acceptance, and usability.

Results

Overview

A total of 31 parents and 22 regional doctors participated in the HTP and received the questionnaire; 25 parents and 15 regional doctors responded (response rate of respectively 81% and 68%). Reasons for not responding on the questionnaire are unknown. Parent and regional doctor characteristics are shown in Table 2.

At the start of treatment, all children were ≤5 months of age. All parents and doctors proved themselves experienced with use of the computer and Internet. Each regional doctor treated an average of 1.5 patients (range 1-4). The mean distance from parents' residence to the tertiary expert center was 52 kilometers.

Table 2. Characteristics of the parents and regional doctors.

Characteristics	Parents, n=25	Regional doctors, n=15
Male-female ratio	1:7.3	1:1.5
Age of respondent, years		
Mean (SD)	32.1 (4.1)	44.9 (9.4)
Median (range)	32.0 (25-41)	39.0 (36-62)
Medical specialty, n/N (%)		
Dermatologist		7/15 (47)
Pediatrician		8/15 (53)
Level of education, n/N (%)		
Low	0/25 (0)	
Moderate	6/25 (24)	
High	18/25 (72)	
Unknown	1/25 (4)	
Indication for treatment, n/N (%) [a]		
Cosmetic	9/25 (36)	
Functional	14/25 (56)	
Ulceration	5/25 (20)	

[a]In some cases there were multiple indications for treatment.

Use

All parents and doctors (regional and academic doctors) used the HTP. The e-consult function of the HTP was used by all parents. The average number of e-consultations was 0.5 per parent/month. E-consultations were mostly sent to (and answered by) the academic doctor, 106/112 (95%) messages, within an average time of 2 days. All parents found that their e-consultations were answered adequately.

Eight regional doctors contacted the academic doctor. Of the 8 regional doctors, 7 (88%) found the academic doctor easily accessible for consultation and questions; 40% (6/15) of the regional doctors used tertiary teledermatology to contact the academic doctor. Others contacted the academic doctor by email or phone. Four of the 15 regional doctors (27%) used the e-consult function to communicate with parents. Some contact moments between doctors and patients were not reported in the HTP, whereas doctors did report in the hospital's own electronic patient dossier.

The different functions of the HTP (e-consult, uploading photographs, completing questionnaires) were used by 19/25 (76%, range 32-100%) of the parents.

Many of the parents (63%, range 52-76%) agreed that treatment by the regional doctor corresponded with the information given by academic center. Parents saved time and costs because of treatment at a regional doctor in respectively 58% and 48% of the cases.

Of the regional doctors, 93% (14/15) felt informed enough to treat patients with IH.

Acceptance and Usability

Perceived Usefulness

A majority of the parents (24/25, 96%) and regional doctors (13/15, 87%) considered the eHealth intervention useful in the care for IH. Parents agreed that different functions (e-consult, PHR, etc) of the HTP were useful (average 21.5/25, 86%, range 84-88%). The different functions of the HTP (eg, IH treatment protocol, access to information about side effects and photographs prior to consultation) were useful according to an average of 87% (range 80-93%) of the regional doctors. There were 7/13 (54%) regional doctors and 22/25 (88%) parents that thought the e-consult function to contact each other was useful. The tertiary teledermatology was thought to be useful by many of the regional doctors who used tertiary teledermatology (75%, 9/12).

Perceived Ease of Use

Instructions on the HTP and patient information were clear according to 92% (23/25) and 96% (24/25) of the parents respectively. Instructions on the HTP and the IH treatment protocol were clear according to 73% (11/15) and 80% (12/15) of the regional doctors respectively. Many of the parents (76%, 19/25) and just over half (53%, 8/15) of the regional doctors agreed on the statement that the HTP is easy to use. Technical problems using the HTP were reported by 28% (7/25) of the parents and 73% (11/15) of the doctors.

Attitude

Most of the parents (88%, 22/25) felt positive about treatment at a regional doctor, and many parents (68%, 17/25) found that treatment at a regional doctor felt safe. All 15 regional doctors (100%) felt positive about transition of treatment from the

tertiary expert center to them, and 92% (23/25) of the parents and 93% (14/15) of the regional doctors felt positive about usage of the HTP. Almost all parents (96%, 24/25) found the HTP was worth the time investment. Although, with 47% agreement, regional doctors reported that they have difficulties with the time investment in the HTP (documenting in the PHR and answering e-consultations). Finally, 72% (18/25) of parents felt more involved in treatment due to the HTP.

The average satisfaction rates parents and regional doctors gave the eHealth intervention on a 0-10 scale were 7.7 (SD 0.75).and 7.3 (SD 1.4), respectively. An overview of the main results is given in Table 3.

Comments and suggestions were evaluated. Positive comments of the parents were given about improvement of access to health care professionals and saving time. Positive comments of the regional doctors included improvement of contact between parents and experts. Points of attention of parents were privacy issues and lack of trust in expertise of regional doctors. Regional doctors were concerned about time investment.

Table 3. Overview of the feasibility of the eHealth intervention sorted by variable.

Feasibility	Parents	n/N (%)	Regional doctors	n/N (%)
Use	Use of different functions (e-consult, uploading photographs, questionnaires) (mean)	19/25 (76) range 32-100	Use of different functions (tertiary teledermatology)	6/15 (40)
	E-consults were adequately answered	25/25 (100)	Informed enough about IH care	14/15 (93)
	Treatment at regional doctor corresponds with information given by academic center (mean)	15.7/25 (63) range 52-76	Easy contact with tertiary caretaker	7/8 (88)
Perceived usefulness	HTP is useful	24/25 (96)	HTP is useful	13/15 (87)
	Usefulness of different functions	21.5/25 (86) range 84-88	Usefulness of different functions (mean)	12.7/15 and 13/14 (87, range 80-93)
	Usefulness of e-consult	22/25 (88)	Usefulness of tertiary teledermatology	9/12 (75)
			Usefulness of e-consultation	7/13 (54)
Perceived ease of use	Instructions of the HTP	23.5/25 (94) range 92-96	Instructions of the HTP	11.5/15 (77) range 73-80
	The HTP is easy to use	19/25 (76)	The HTP is easy to use	8/15 (53)
	Technical problems	7/25 (28)	Technical problems	11/15 (73)
Attitude	Positive about treatment at secondary caretaker	22/25 (88)	Positive about treatment at secondary caretaker	15/15 (100)
	Positive about usage of HTP	23/25 (92)	Positive about usage of HTP	14/15 (93)
	Worth the time investment	24/25 (96)	Worth the time investment	7/15 (47)
	More involved in care	18/25 (72)		

Discussion

Principal Findings

This study describes an eHealth intervention to make the care for children with IH efficient and easily accessible using an online hemangioma treatment plan. Treatment of IH took place with regional doctors, supported by an expert at distance (academic doctor). The HTP was used to facilitate transition of treatment to regional doctors and to involve parents in the care for IH. Evaluation of the feasibility of this new way of providing care was performed by studying the small group of first patients and regional doctors who participated in this newly developed eHealth intervention.

The feasibility according to the parents ranged from 63-100% and according to the regional doctors from 47-100% (Table 3). The predetermined percentage of 90%, necessary to qualify the eHealth intervention as feasible, was not always reached. However, almost all parents thought the HTP was useful and all regional doctors had a positive attitude towards the HTP.

Although most results on feasibility are positive, only 53% of the regional doctors found the HTP easy to use. This could be influenced by the fact that 73% of them experienced technical problems and that they were mostly instructed in writing. Most technical problems experienced by both parents and doctors related to logging in and uploading of photographs. The problems were most likely caused by prematurity of the technology itself. Problems of logging in are resolved now. The problem with uploading photographs was due to low capacity of the website and will be resolved in the near future. Technical problems might also be caused by a lack of adequate computer skills to use eHealth systems. Health care providers are the key driving force in pushing eHealth initiatives [18]. IT support (verbal) might facilitate eHealth acceptance and use [19].

Furthermore, the tertiary teledermatology to consult an academic doctor of the CAVU team was seldom used by the regional doctors. Mostly they consulted the CAVU team by phone or by sending emails. Probably the regional doctors were not used to this tool for consulting a colleague, and the fact that parents could see the content of the questions of the regional doctor may have contributed. However, studies have shown that tertiary teledermatology might improve communication between regional and academic doctors and might reduce wait times [20,21]. They have advantages over telephone consultations, which require the need for both parties to be available at the same time, and email, which does not meet current privacy requirements for sharing personal health information [20,22].

Secondly, regional doctors were not positive about the e-consult with the parents. Only about half (54%, 7/13) of regional doctors considered the e-consultation useful. A possible explanation is that these doctors might expect e-consultations to be time consuming (74% of the regional doctors found the HTP worth the time investment) or that they are not used to working with e-consultations. However, it has been shown that the use of e-consultations in dermatology is feasible and the majority of e-consultations take less than 10 minutes for the medical doctor to answer [20,23,24]. It has been shown that e-consultations improve access to specialty care [25]. On the other hand, Palen et al [26] found that having online access to medical records and clinicians was associated with increased use of clinical services. However, this has been debated by others [27,28]. Further research should point out the consequences of e-consultations for time investment and usual care.

Some parents felt that the regional doctor was less experienced with IH care. However, in our opinion, this is probably only a temporary problem since studies have demonstrated that online health communities of doctors from different echelons and patients can be used to exchange medical experience and knowledge and that knowledge of participants increased and the adherence to guideline recommendations improved [29,30]. Partial transition of treatment of IH to secondary centers combined with support by an expert might have an educational value for the regional doctors. This is confirmed by the fact that 87% of the regional doctors agreed that the HTP was of educational value for them. In the long term, increased knowledge about IH treatment and treatment indications might result in better recognition and treatment of children with IH at risk for complications. In the short term, increasing the knowledge about treatment of IH among regional doctors (eg, through eLearning courses) might make them and the parents more comfortable with treatment in regional centers.

Some of the parents included in this study mentioned privacy issues. Security and privacy issues are consistently found in studies as influencing patients' interest in digital PHRs [31]. Adequate verbal instruction to parents about (the prematurity of) the intervention and about security is important in further implementation. The implementation of the HTP requires a different way of acting and thinking from both doctor and patient. Satisfaction of eHealth interventions has rarely been studied [32]. High patient satisfaction was also seen in a Dutch eHealth intervention that includes an eczema portal combining e-consulting, monitoring, and self-management training for patients and parents of young children with atopic dermatitis [33,34]. Overall parents were positive about the eHealth intervention. From the perspective of the regional doctor, feasibility of treatment at local hospitals and system usability of the HTP should be further adapted to their needs to enhance acceptation, actual usage of the HTP, and implementation on a larger scale. It is known that medical doctors have had some reservations about moving forward in the area of eHealth and PHRs, partly because of concern that they will be bombarded with questions and that patients will have trouble interpreting their findings [35,36]. However, most of the empirical experiences suggest that these problems do not represent major issues when patients are provided with and adopt PHRs [37,38]. Solutions to reduce time investment could involve specialized nurses in the triage of e-consultations and creating a link between the patient file of the hospital and the PHR of the eHealth intervention. Nevertheless, results of this early evaluation of the HTP should be interpreted with consideration of the psychology that goes along with changes in management. Hands-on training for the set-up could be necessary for structural implementation of the e-consultation functionality [25]. Besides patient-doctor interaction, workforce items such as workload and workflow, as well as contextual factors like institutional policy regarding eHealth, influence the implementation of eHealth interventions [39]. To realize treatment of IH on a larger basis at local hospitals, clear referral policy should be made. Regional doctors should agree on how to facilitate treatment of IH on a larger scale. Academic doctors have an important role in assuring the quality of care as they are expected to recognize those patients that require treatment at the tertiary center. Hospital management of tertiary centers should incorporate e-consulting in daily practice to ensure the academic doctor can meet this important role.

Limitations

The results of our study must be interpreted with caution given the small sample size of both parents and doctors and the prematurity of the intervention. Furthermore, parents had a relatively high education level and were therefore not representative of the general population.

The implementation of eHealth interventions will incur costs. However, the transition of IH treatment to secondary centers might save (in)direct health care costs. Health insurance does already reimburse the implementation of eHealth in some fields of medicine. However, there is still no funding for the care provided through the eHealth interventions. Studies have shown that eHealth, combining e-consultations, monitoring, and self-management training, could lead to cost-savings, and e-consultations could reduce the number of face-to-face consultations [20,23,24,40]. In this study, parents reported a time and cost reduction with respect to traveling due to the use of the HTP. Lower costs can be expected due to a lower number of face-to-face consultations at the tertiary academic center. There might even be situations where digital contact through the HTP replaces face-to-face contact. Wait times will shorten, and more new patients can be seen in a shorter time. Lower indirect costs can be expected due to lower work-absenteeism, as care can be received closer to home. Further studies are necessary to confirm this.

Conclusions

The HTP is a new care innovation that was and will be continuously improved according to user feedback. Points for improvement are resolving the technical problems, such as extending the capacity for uploading photographs, providing more detailed training for regional doctors, and taking care of organizational support. The HTP was a pilot to evaluate the feasibility of treating patients in a regional hospital with online support of the academic center and was part of the pediatric Skin House. After some improvement, this eHealth intervention could be a helpful tool for efficient and accessible care for IH and might be used to increase cooperation between different sectors of health care (primary, secondary, and tertiary care).

Our eHealth intervention to improve the efficiency and accessibility of care for children with IH shows good feasibility, especially among parents. Improvement with respect to technical problems, training of regional doctors, and achieving organizational support might be needed for successful implementation in the future.

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Conflicts of Interest

None declared.

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Abbreviations

CAVU: Center for Congenital Vascular Anomalies Utrecht
HTP: hemangioma treatment plan
IH: infantile hemangioma
PEU: perceived ease of use
PHR: personal health record
PU: perceived usefulness
TAM: Technology Acceptance Model

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Protocol

A Formative Evaluation of a Social Media Campaign to Reduce Adolescent Dating Violence

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Abstract

Background: The Emory Jane Fonda Center implemented the Start Strong Atlanta social marketing campaign, “Keep It Strong ATL”, in 2007 to promote the development of healthy adolescent relationships and to foster the prevention of adolescent dating abuse among 11-14 year olds.

Objective: A formative evaluation was conducted to understand whether messages directed at the target audience were relevant to the program’s relationship promotion and violence prevention goals, and whether the “Web 2.0” social media channels of communication (Facebook, Twitter, YouTube, Flickr, Tumblr, and Pinterest) were reaching the intended audience.

Methods: Mixed methodologies included qualitative interviews and a key informant focus group, a cross-sectional survey, and web analytics. Qualitative data were analyzed using constant comparative methodology informed by grounded theory. Descriptive statistics were generated from survey data, and web analytics provided user information and traffic patterns.

Results: Results indicated that the Keep It Strong ATL social marketing campaign was a valuable community resource that had potential for broader scope and greater reach. The evaluation team learned the importance of reaching adolescents through Web 2.0 platforms, and the need for message dissemination via peers. Survey results indicated that Facebook (ranked 6.5 out of 8) was the highest rated social media outlet overall, and exhibited greatest appeal and most frequent visits, yet analytics revealed that only 3.5% of “likes” were from the target audience. These results indicate that the social media campaign is reaching predominantly women (76.5% of viewership) who are outside of the target age range of 11-14 years.

Conclusions: While the social media campaign was successfully launched, the findings indicate the need for a more focused selection of communication channels, timing of media updates to maximize visibility, balancing message tone and delivery, and incorporating differentiated messaging for the target audiences. Collaboration with parents and community partners is also emphasized in order to expand the campaign’s reach and create more channels to disseminate relationship promotion and dating violence prevention messaging to the intended audience.

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KEYWORDS

adolescents; dating violence; social media; formative evaluation

Introduction

Background

Adolescence is a developmental period in a youth's life characterized by biological and physical changes, social role transitions, and experimentation with dating relationships [1]. According to the Youth Risk Behavior Surveillance survey, 10% of adolescent relationships result in physical violence [2]. Specific demographics are more likely to experience dating violence, including African American adolescents [2], and older adolescents (ages 16-18) who have a significantly greater risk of involvement in abusive relationships [3]. Furthermore, particularly among younger adolescents, physical aggression is perpetuated by both sexes [4-7].

Social Networks and Social Media

Social networking websites, termed "Web 2.0," are principal platforms in the current state of the Internet [8]. Such websites provide a global community for individuals to contribute and respond to content [9]. In the United States, 93% of adolescents aged 12-17 access the Internet and 73-80% participate in one or more networking sites [10,11]. However, trends indicate that the youngest adolescents (ages 12-13) utilize social media at significantly lower rates. While 87% of young adolescents and 95% of older adolescents report they have a Facebook profile [12], most social media sites report less than 10% of their users are under 18.

Numerous studies suggest the effectiveness of using social media initiatives to communicate health information [13,14]. Approximately one third of adolescents ages 12-17 (31%) access the Internet for health information [15]; African American youth, girls, and lower income youth are more likely to seek health information on the Internet [14-16]. These data indicate that social media could be a highly effective tool in engaging these higher risk adolescent groups in health communication and promotion.

Formative Evaluation of Start Strong ATL

Start Strong is a national program designed to promote healthy relationships and prevent teen dating violence [17]. One of the program's core objectives was to design and implement communication strategies to promote the program and engage youth. Start Strong Atlanta (ATL) implemented by Emory University's Jane Fonda Center, worked with youth leaders to create and maintain a youth-focused website, striving to reduce teen dating violence among 11-14 year olds (Figure 1) [18].

The purpose of the study was to examine the effectiveness of the campaign during its pilot testing period, which utilized social media platforms as dissemination to reduce teen dating violence, improve healthy adolescent relationships, and promote positive social and cultural norms [19-22]. Specifically, this evaluation sought to examine implementation challenges related to message dissemination via selected communication channels including Facebook, Twitter, YouTube, Flickr, Tumblr, and Pinterest (Figure 2) [20,23,24]. Given the lack of precedent in developing such social media campaigns, the evaluation focused on issues related to message receptiveness, source evaluation, and behavioral changes as a result of message exposure. The lack of precedent in developing social media campaigns is known for teen dating violence, but may be a more generalized phenomenon. Therefore, the results from this study will have a broader impact on the message framing and content for future online campaigns. The recommendations from this study will also aid in the future to develop successful methodologies of tailored campaigns for the young adolescent target audience in order to expand reach and awareness [25].

Figure 1. Start Strong ATL program website.

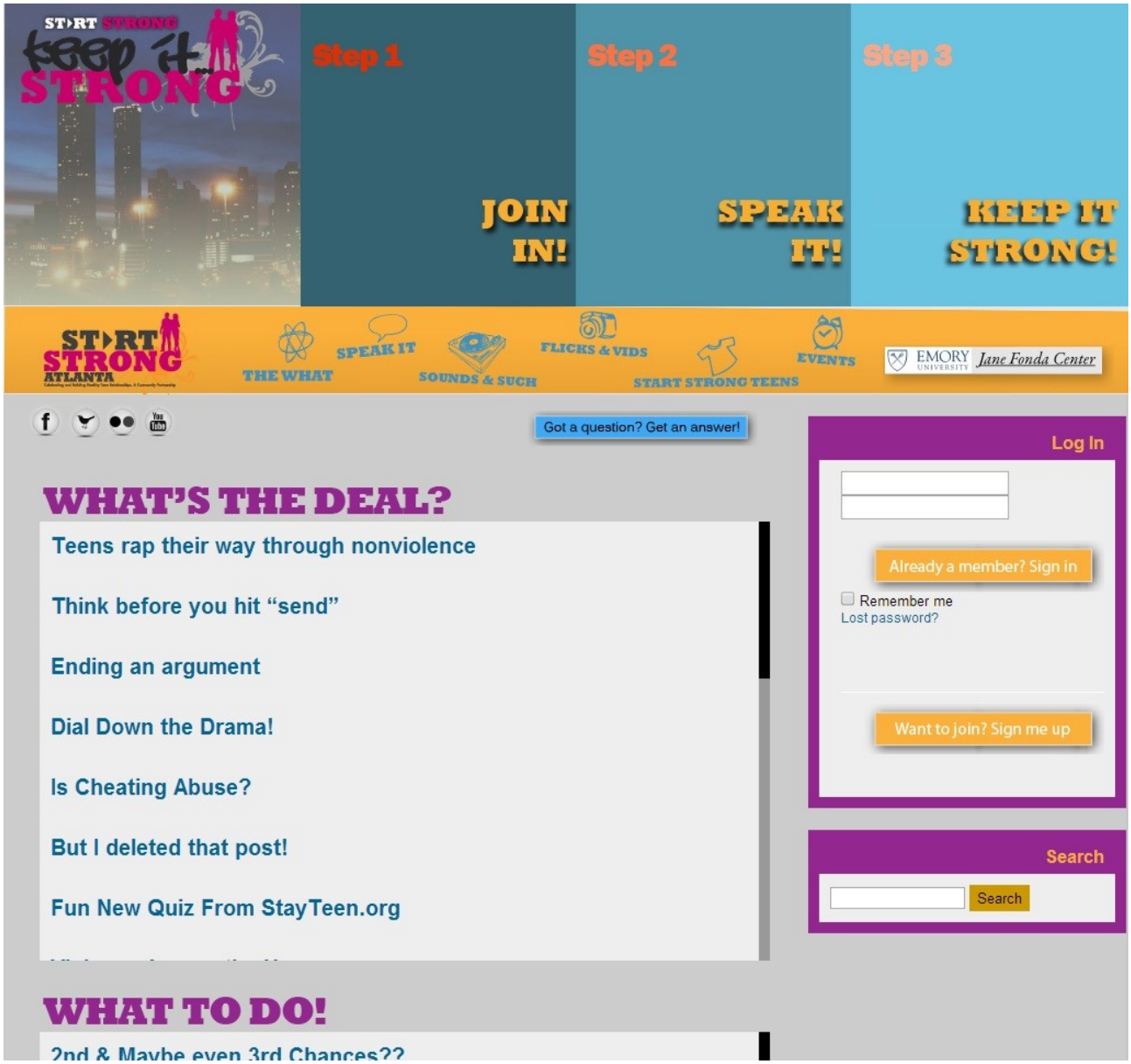
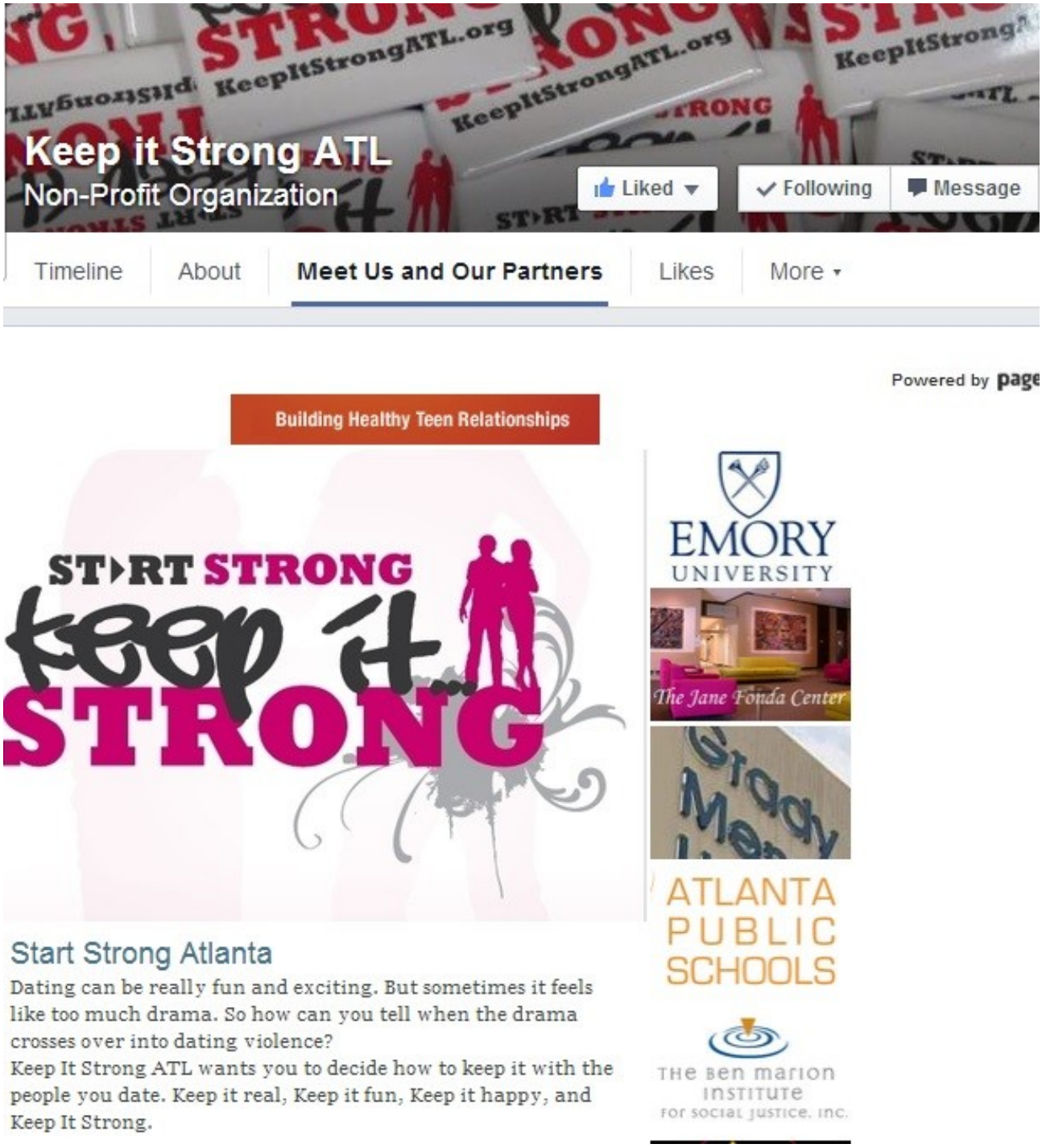


Figure 2. Start Strong ATL corporate Facebook page.



Methods

Study Recruitment

The current and previous full-time staff members (n=4) ≥18 years of age who were responsible for developing and implementing the social media campaign were recruited in person and via email from the Start Strong ATL program headquarters. These four staff members represent the medical director, project manager, former communications/media coordinator, and current communications/media coordinator. Current Start Strong ATL teen leaders (n=8) who serve as peer educators, mentors, and contributors for social media content were recruited through the Jane Fonda Center via email. Key informants, specialized in adolescent health and health communication, conducting similar social media campaigns (n=3) were recruited via email from Planned Parenthood Southeast (SE) and Emory University's Office of Health Promotion.

A combined total of 15 respondents participated in qualitative interviews or a focus group. All 15 respondents were asked to complete an additional quantitative survey upon completing their interview. Of these respondents, 12 completed the additional quantitative survey. Google analytics were collected to examine traffic and user behavior from the Start Strong ATL program website from 2010-2012. A total of 2929 unique users

from the program website were included in the analysis. Facebook Insights were also examined to isolate trends in user behavior across both the Start Strong ATL Facebook and Twitter page, which were linked to allow for simultaneous postings.

Evaluators collected data between August and December 2012, garnering comprehensive information from program staff and youth leaders, and communications personnel from community organizations. Discrete populations of respondents were used for data collection, such that all Start Strong ATL teen leaders interviewed were independent of the staff and web designers utilized to build the campaign's website and social media sites. Additionally, all key informants interviewed were in no way contributors for or associated with the Start Strong ATL social media campaign. Participants were required to provide verbal informed consent prior to their participation. All participants were interviewed at their respective organizations to ease participant burden.

Formative research typically involves gathering data from a small number of stakeholders as the purpose of this type of evaluation is to provide insight into social media content, deployment, and use potential or its feasibility [26]. With a focus on development, this type of evaluation for social media campaigns often relies on those methods that produce results with the least amount of participant burden (eg, real-time third-party tracking data). In practice, formative research may include very small numbers of participants for primary data collection via interviews, focus groups, or surveys as a program is being created [27,28]. Capturing an adequate number of respondents needed to answer "pilot phase" social media campaigns such as Start Strong, is also weighed against the commonly held evaluation standards of feasibility, utility, propriety, and accuracy of any resulting information from the investigation [29,30].

Data Collection

The evaluators implemented a mixed methods assessment using qualitative interviews and quantitative surveys. Semi-structured interview and discussion guides were utilized to capture qualitative data; data were audiotaped and subsequently transcribed verbatim by team members. Participant burden was restricted to 45-60 minutes for key informants, and 60-90 minutes for in-depth interviewees.

The survey was developed to garner quantifiable data about message receptiveness, source evaluation, and potential behavioral outcomes of the Keep It Strong ATL social media campaign. The survey consisted of 15 items, including 2 demographic questions indicating age and gender, and 2 open-ended qualitative questions. Participants completed the questionnaire in 10 minutes or fewer to alleviate burden.

Analytic Strategy

The social marketing principles of product, promotion, price, and place informed the development of focus group and interview questions on the use of planned social media and its content on adolescent dating violence. Drawing upon similar formative studies, the evaluators explored questions related to the campaign (product), how it was being disseminated among target audiences (promotion), the opportunity cost of time spent online and with content (price), and the utility and feasibility of specific online venues for teen dating violence content [31-34].

Evaluators used the constant comparative method, informed by grounded theory, for qualitative analysis. Evaluators continuously compared emerging themes, concepts, and indicators [35]. Two evaluators coded each interview independently before both sets of codes were compared to assess inter-coder reliability. All coding achieved a Cohen's kappa statistic of ≥.8 indicating a satisfactory level of agreement between the raters [36]. Emergent themes with an agreement threshold of ≥.8 among all respondents were classified as major themes. Only major themes, indicating commonality among the informants, were selected for inclusion in the results.

Quantitative data were analyzed with SPSS 20.0 to examine frequencies and assess the perceived effectiveness, reach, and adherence to the program and its objectives. Open-ended questions were compiled, coded, and analyzed according to previously stated parameters. Descriptive statistics and corresponding graphics were generated for all variables of interest.

Google analytics were also utilized to conduct oversight of website traffic since live production in November 2010 to analysis in November 2012. The software collected broad demographic information concerning users, frequency and duration of visits, and mechanisms to gain traffic. Additionally, evaluators analyzed Facebook Insights, which recorded all demographics and traffic-related information for the corporate Keep It Strong ATL Facebook page. From this source, evaluators generated descriptive tracking statistics and corresponding graphics to understand the page's reach, specifically unique viewers of original content and "viral" spread to subsequent users.

Results

Summary of Results

Data indicate that the Start Strong program was effective in educating adolescents and adults about reducing dating violence and promoting healthy relationships. On average, adolescents ranked the program at 8.25 (out of 10) for reducing violence and at 8.67 for promoting healthy relationships, whereas adults' responses averaged 7.83 for both categories. Data were compared across all collection methodologies, and major themes, with an agreement threshold among respondents of ≥0.8, were classified. The major themes addressed in this paper are Communication Channels: Technology, Timing, and Refreshing Social Media; Message Source: Tone; and Message Framing: Prevention Messaging and Content. These results were pervasive among qualitative in-depth interviewees, survey results, focus group participants, and web analytics, and were the basis for informing implications for future research.

Characteristics of the Sample

The sample is an aggregate representation of web users, key informants from the community, teen leaders, and current and former Start Strong program staff. Broad demographic data were generated for web users (n=2929) via Google analytics

and Facebook Insights. The website data indicated 43.8% of the total users were between 18 and 24 years old, and over half were female (68.8%). In-depth qualitative interviews, focus group, and quantitative survey were administered to individuals affiliated with Start Strong ATL and the broader Atlanta adolescent health promotion community, including youth leaders (n=8), key informants (n=3), and program staff members (n=4). The demographic most represented is the 15-24 age cohort (age ranges included 11-14, 15-24, 24-35, and ≥35 years), and the sample consisted of marginally more females (n=8) than males (n=7).

Communication Channels: Technology, Timing, and Refreshing Social Media

Adolescents in the focus group expressed that they are consumers of technology and social media, particularly Twitter, Facebook, and Instagram, and recommended that the campaign continue to disseminate information to adolescents via these outlets. One youth leader stated:

> *Since it's social media, and technology is like the world, since (sic) everyone interacts through social media, I think that's a big door for teenagers to get that type of information.* [Ashlee]

Social media is an effective vehicle for sharing information with adolescents who use social media for daily interactions. Twitter and Facebook were the most frequently visited sites; the majority of sampled participants viewed them between daily and weekly.

Participants also demonstrated that Facebook and Twitter hold the greatest appeal for adolescents aged 11-14 (ranked 6.5 and 6.09 out of 8 respectively). However, Keep It Strong ATL maintains two Facebook pages: personal and corporate accounts. The personal page was ranked at 6.5 while the corporate account was ranked at 4.33. The pages with the least appeal were the Pinterest page, ranked at 3.75, and Flickr, ranked at 3.5. The program webpage was ranked at 4.82 (Figure 3).

Communication dissemination challenges, which minimized the sites' ability to reach the target audience, were also identified. Key informants emphasized that awareness of adolescents' social media usage is important when planning updates and posts on social media sites to achieve a high level of visibility. Carefully timing updates optimizes the target audience's engagement. One key informant explained:

> *...on a Sunday night, that's a pretty common time for people to be on Facebook, or on Twitter…between the hours of like 8:00pm and 11:00pm, you know because people are trying to procrastinate from their homework that they have due on Monday… start that hype at night when people are more likely to be on there.* [Michael]

Hence, key messages and programmatic content may be most effective when posted at night.

Analytics reflected the participants' sentiments and provided insight into the program website traffic and engagement. From early November 2010 to late November 2012 the program's website generated 2929 unique visitors and 9618 page views, of which 866 people visited the website multiple times (Figure 4). The average duration of each visit was 2 minutes, with 2.55 pages viewed per visit, which indicates quick page turnover among viewers. Most traffic, 77.05%, was generated by first time visitors, indicating continuous efforts to promote dissemination, but also potentially indicating that the content grew stale, given that the majority of visitors did not return to engage with updated posts. Further examination of daily visits from 2010-2012 revealed alternating periods of traffic spikes and lows (Figure 5). During initial stages of implementation, views steadily increased, followed by a sharp spike corresponding with a Public Service Announcement (PSA) video contest. Views subsequently dropped off until October 2011 when staffing changes were made. Traffic noticeably increased after this point, as the website URL was repeatedly intertwined with Facebook and Twitter postings. The most views ever reached in one day was 27, a peak which coincided with the PSA video contest; views only exceeded 20 on five additional occasions over the two years. Quantitative Facebook Insight data revealed that 66.1% of users who "like" this page were females, outside the target audience of 11-14 year olds; only 3.5% of "likes" were from the target audience (Figure 6).

Figure 3. Social media site appeal for adolescents and adults as reported by program staff, (N=12).

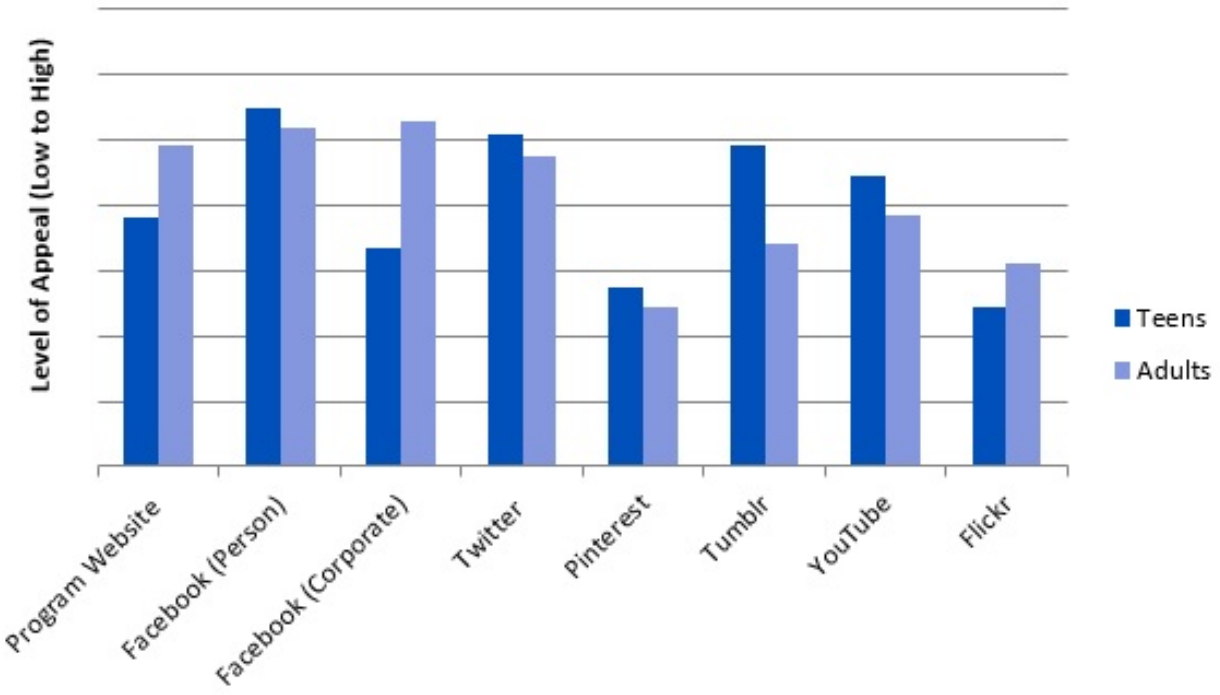


Figure 4. Quantity and frequency of the Start Strong website visits, 2010-2012, generated in Google Analytics.

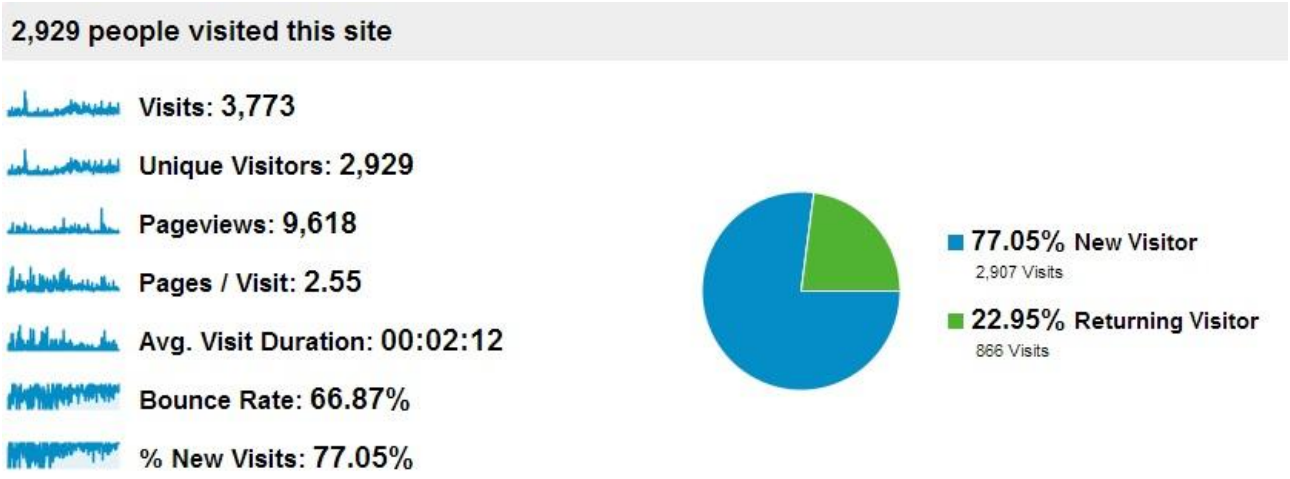


Figure 5. Frequency of daily visits from the Start Strong website, 2010-2012, generated in Google Analytics.

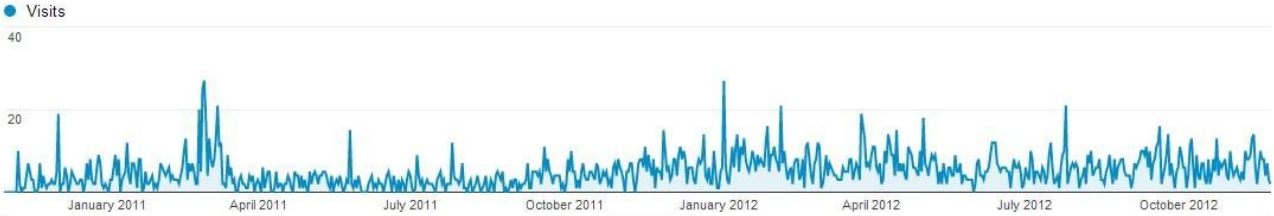


Figure 6. Age distribution of Facebook Likes on the Keep It Strong ATL corporate website, generated in Facebook Insights (n=223).

Message Source: Tone

The Keep It Strong ATL social media campaign was uniquely focused on positive messaging. Staff felt that negative content was prominent on media outlets, thus exclusively positive content was posted on program-run sites. Key informants were largely in agreement about the importance of positive messaging. However, one key informant stated:

> *I believe very strongly in presenting positive images and role modeling… around creating a world without violence, but saying that's our only focus would be really diminishing of the students who need support.* [Lauren]

Social media users who have been victims of dating violence may need additional support. Another key informant mentioned that posted messages with the most views, "likes," and "shares," were humorous. She said:

> *Things targeting teens, we want to be serious in our messaging, but we don't want to come off as being strict or stringent or anything, just kind of humor, a little sarcasm.* [Sara]

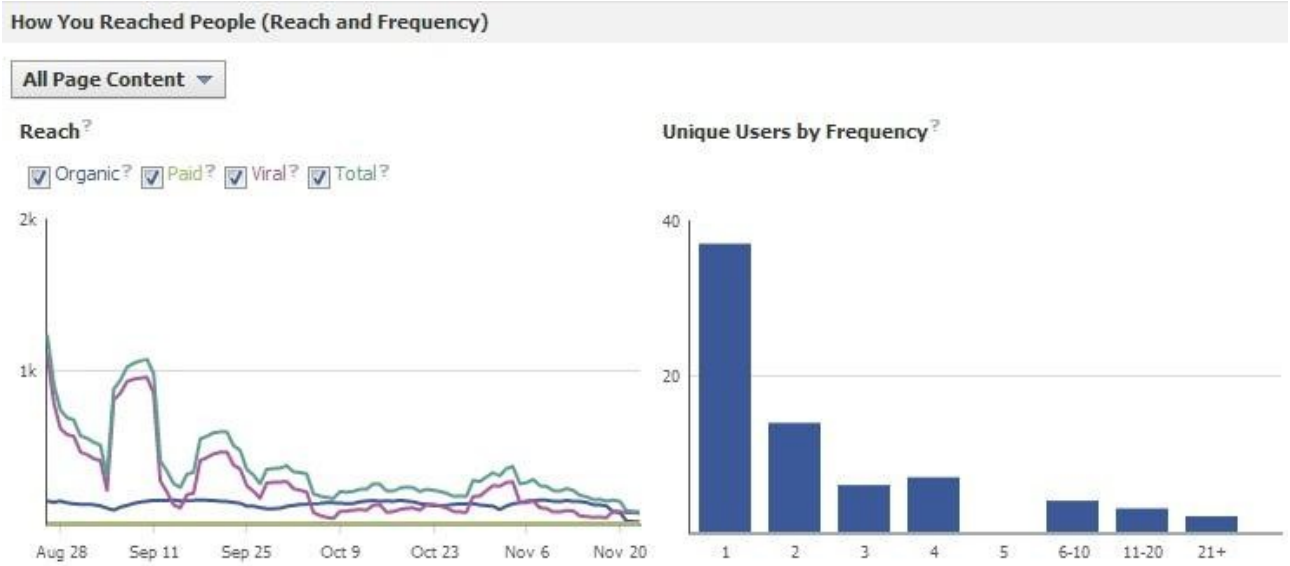
Teens respond to sarcasm and humor since they often use it in communication with each other.

Another key informant highlighted the realities of using social media to convey educational messages. The most effective tone is communicated quickly and efficiently, in short bursts. Bursts effectively gain attention without inundating with information, as this informant states:

> *It's something to start awareness and hopefully gain someone's attention in some way. And once you have them as an audience, then hopefully you can educate them after. But you're not going to solve [dating] violence using Facebook or Twitter.* [Vanessa]

Analytics highlight the results of this strategy. The site's reach steadily declined from over 1000 users to under 200 within a 3-month span. There was one period of increased reach, corresponding to when Facebook posts were linked with Twitter, but when simultaneous posting ceased, reach began to rapidly decline (Figure 7).

Figure 7. Frequency and quantity of reach per unique views for the Corporate Facebook page, generated in Facebook Insights.



Message Framing: Prevention Messaging and Content

Key informants focused on prevention and preventive education, but also stated that prevention should not be the only campaign aim as demonstrated:

> *we can't only focus on prevention, we can't only talk about aspirations toward what healthy relationships look like, and consent… we do need to talk about students' stories and experiences, we do need to talk about resources…that involves us working…on multiple levels for prevention.* [Lauren]

This suggests that primary prevention would be more effective in combination with secondary and tertiary prevention efforts, to support those who have experienced or witnessed adolescent dating violence. Stratification of user tracking data revealed limited amounts of "sharing" and reposting of Keep It Strong ATL corporate webpage and Facebook content. Females, aged 18-34 years, were the greatest proportion (59.4%) of those who reposted or shared content. This group likely found the topics of adolescent dating violence and healthy relationships relevant, motivating their engagement with the conveyed messages.

Discussion

Principal Findings

This formative evaluation revealed critical challenges and opportunity for the Keep It Strong ATL social media campaign [37]. During its pilot phase, the campaign was successful with engaging women and persons over 18 years of age. Yet, the data indicate that it did not reach or maintain the interest of the target audience, those in the 11-14 year old age range who remain vulnerable to adolescent dating violence [4,15]. In particular, results from qualitative and quantitative data reveal the importance of optimizing the capability of communication channels, placing emphasis on message framing and content, and selecting the right messengers or sources for success [19]. Foremost of concern for this audience is inclusion of appropriate technology, timing message dissemination, and refreshing social media content to maintain visibility and interest among adolescent viewers [15,16,19].

The results indicated that the campaign's use of Facebook was the campaign's most effective platform in generating viewers [38,39], particularly paired with YouTube self-generated content, links from partner pages, or relevant videos [40]; however, the majority of viewers were not representative of the target audience. Findings also indicate the importance of updating media sites during evening and weekend hours when traffic is highest and the targeted young adolescents are not in school. Yet, caution must be exercised as previous studies have linked social media use patterns to poor child sleep and health outcomes [25,41]. Adolescents in this study also emphasized the importance of timing and refreshing of messages, and improving visual messaging to promote rapid dissemination among the target audience [42]. Similar approaches have been taken with social media to reduce stigma through increased social support generated for those living with HIV in Nigeria [43]. Thus, consideration of appropriate technology use, timing of message delivery, and maintaining content currency will improve social media campaign objectives for adolescents [43,44].

Backed by Prospect Theory [45,46] and developed by psychologists Amos Tversky and Daniel Kahneman [47,48] message framing has been proposed as a potential method to promote positive health-seeking behavior [49]. Message framing in health communication typically recognizes that individuals tend to avoid risks when considering gains and prefer risks when considering losses. In this framework, gain-framed messages are developed to communicate information by emphasizing the benefits of the target health behavior. Loss-framed messages are created to emphasize the risks of not engaging in a behavior [50-54]. The campaign's use of positively-oriented "gain-frame" messages communicate violence prevention information, emphasizing building and maintaining healthy relationships [50]. Furthermore, meta-analytic evidence from a variety of investigations indicate that gain-frame messages may be more persuasive in promoting preventive health behaviors (eg, avoiding negative relationships) [50-55]. However, results also suggest the need for message framing improvements. Positive messaging can and should remain a central focus; however, youth require context, which entails providing examples of unhealthy or inappropriate behaviors as a point of discussion and reference [5,56,57]. Furthermore, it is necessary to include resources for individuals who may have already experienced abuse or unhealthy relationships [5].

Parents and guardians often impose restrictions on preteens' access to social media sites during this developmental stage [15]. Results indicate that the Keep It Strong ATL social media campaign did not reach its intended target audience of 11-14 year old adolescents. We believe this may be due to a lack of awareness of the existence of the program and Facebook sites, access restrictions, and messages and content that did not engage the target audience. Thus, reaching parents and other adults, such as counselors and teachers, via a theory-driven campaign approach represents an opportunity to engage the younger target audience. For example, the "Parents Speak Up National Campaign" (PSUNC) was a theory-driven multimedia parent-child campaign that achieved positive normative and outcome expectancy shifts by targeting both parents and adolescents in a "wait until older" message strategy [58]. Similarly, the Centers for Disease Control and Prevention's (CDC) multipronged VERB media campaign targeted parents of children ages 9 to 13 years and achieved positive outcomes related to physical activity attitudes, beliefs, and behaviors which corresponded with greater than 50% of parental awareness of VERB by its third year of inception [59,60]. The formative work guiding message development, delivery, and targeting dyadic parent-child audiences evidences successful strategies that can be emulated for adolescent dating violence campaigns.

Additionally, evaluation results indicated that the program would benefit from an "Integrated Marketing Communication" (IMC) strategy that incorporates social marketing into the campaign [61]. When IMC is utilized, the campaign has greater potential to leverage the online power of schools and the broader community to increase the campaign's relevancy and its reach to parents and adolescents. Drawing upon community capacity theory, the IMC approach has greater potential to engender

support from an array of community partners meaningfully connected to the issue [62,63]. Such an opportunity presents the campaign with the greater potential to utilize all elements necessary for its success - appropriate communication channels, message sources, and salient content - to achieve program objectives.

Limitations

We acknowledge that the sampling of a small group of participants from one southeastern city is not representative of eleven other Start Strong cities. Additionally, we recognize the potential for participatory bias, as those who agreed to participate were included and therefore may not be representative of the actual campaign population, partner organization staff, and target audiences. As this evaluation specifically examined a health campaign implemented virtually, limitations of technology must be considered. The Keep It Strong ATL social media campaign platform restricted content mapping onto the existing format, so content often grew stale and outdated, which did not encourage repeat visitors. A greater technological implication was the potential limiting impact of media sites' policies and regulations concerning use of those in the 11-14 year old target audience. Those from the 11-14 year age range could have potentially entered older ages (invalid birthdates) to establish their accounts and bypass policies of accessibility due to age. Hence, absolute conclusions cannot be drawn about audience behavior.

Conclusion

The Keep It Strong ATL social media campaign was successful in launching its website and Facebook sites to achieve audience engagement. Yet, results indicate the need for focused selection of communication channels, improved and resonant messaging for target audiences, and the importance of working with parents and communities to achieve broader campaign dissemination among those most vulnerable to adolescent dating violence.

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Conflicts of Interest

None declared.

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Abbreviations

CDC: Centers for Disease Control and Prevention
IMC: integrated marketing communication
PSA: public service announcement
PSUNC: Parents Speak Up National Campaign
SE: Southeast

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Original Paper

An Online Health Community for Aneurysmal Subarachnoid Hemorrhage Patients: A Pilot Study

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Abstract

Background: Aneurysmal subarachnoid hemorrhage (aSAH) is a condition affecting relatively young patients and has high rates of morbidity and mortality. Online health communities have emerged to fill the void for patient advocacy and information, allowing individuals with shared experiences and chronic disorders to connect.

Objective: We have developed an online health community for aSAH patients, and this pilot study was conducted to evaluate it from a patient's perspective.

Methods: We implemented an online, members-only, health community (MijnSAB, translation: MySAH) in addition to the usual aSAH care at Radboudumc, Nijmegen, the Netherlands. A questionnaire that was sent to consecutive aSAH patients was used to evaluate the usability and utility of MySAH. Answers were provided using a 5-point Likert scale. There was also one open-ended question asking about what was missing from the MySAH tool.

Results: In total, 66 consecutive patients with aneurysmal subarachnoid hemorrhage were informed about the online health community. Of 64 potential MySAH users, 26 patients gained access to MySAH, 20 of whom were willing to participate in the evaluation. Those who used the community were younger (P=.03) and in a better condition at discharge (P=.03). The patients were positive about MySAH's contribution to the quality of their care, but not to their quality of life. Most patients (18/20, 90%) reported that they would recommend the community to others in their position. Open suggestions on how to improve the tool included more frequent blogs, including by a rehabilitation specialist.

Conclusions: This pilot study showed that the online health community, MySAH, has a beneficial effect on the aftercare of patients suffering from aSAH because it gives easy access to relevant information provided by peers or caregivers. Due to the variable clinical outcomes after aSAH, the tool will mainly be useful for a select group of patients (with a better clinical outcome).

(JMIR Res Protoc 2014;3(4):e60) doi:10.2196/resprot.3736

KEYWORDS

subarachnoid hemorrhage; online community; quality of care

Introduction

The incidence of aneurysmal subarachnoid hemorrhage (aSAH) is approximately nine cases per 100,000 [1]. The condition affects relatively young patients, with an average age at first onset of 55 years, and has significant rates of morbidity and mortality [1]. Health-related quality of life is also significantly reduced compared to the normal population [2-4]. Online health communities are increasingly being used to assist these patients and can be a valuable addition to the standard clinical and outpatient care [5-7]. Such communities give patients the opportunity to communicate with professionals and peers and to learn more about their disease and future expectations [8].

We have developed an online health community for aSAH-patients, and this pilot study was conducted to evaluate it from a patient's perspective.

Methods

Implementation

We implemented an online, members-only health community (MijnSAB, translation: MySAH) as an addition to the usual aSAH care provided at Radboudumc. The tool has three main functionalities, which are in line with those described for other online communities [5]. First, information on relevant news can be provided in blogs. Second, the resource is an interactive forum whereby patients can contact others with the disease or put questions to the medical team. Third, general information concerning several aspects of the disease is provided. An example of the access page in the form of a poster used for promotion purposes among patients is shown in Figure 1. Examples of the translated content ("Can I?") are set out in Multimedia Appendix 1.

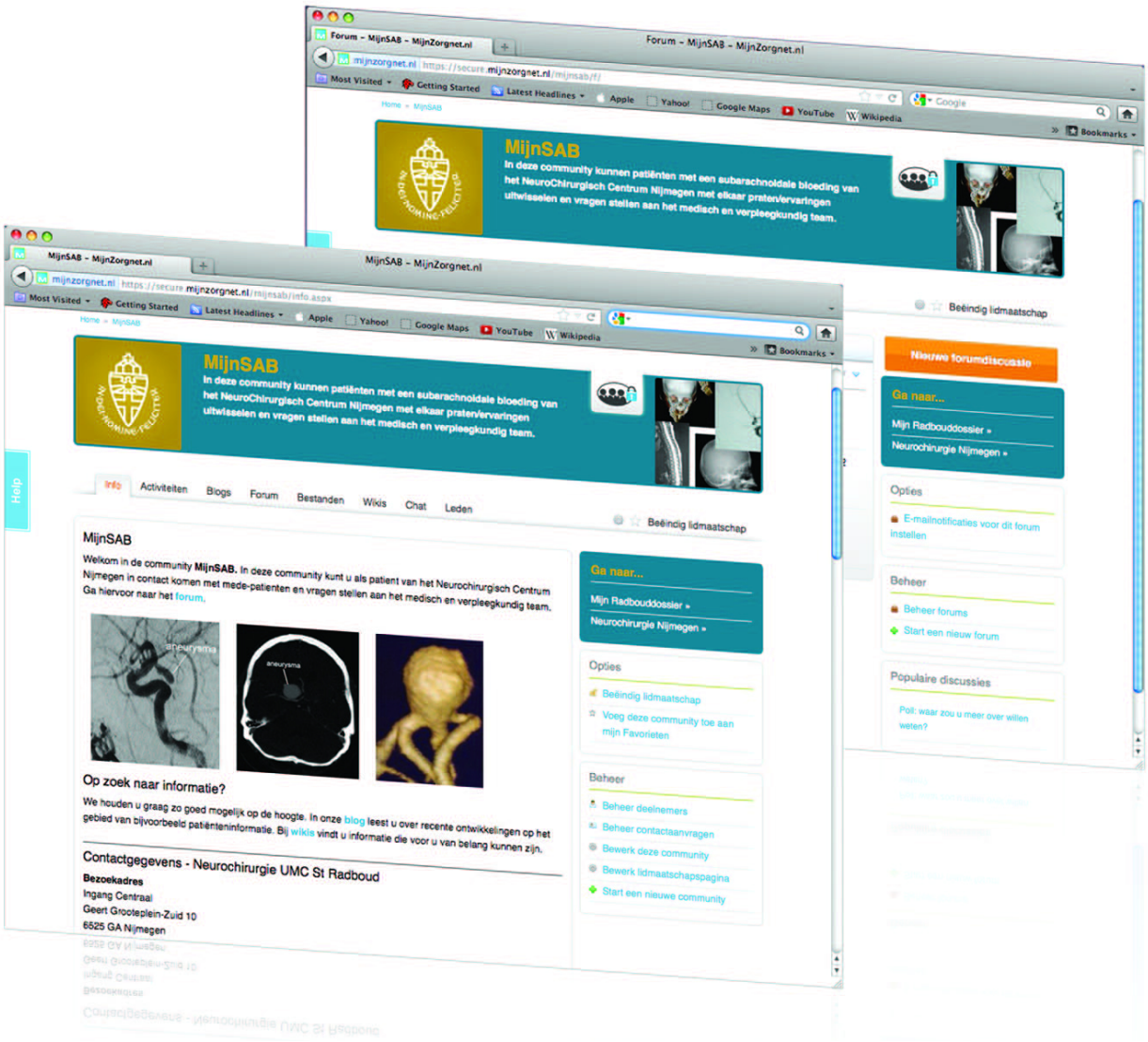
The community tool has been used by Radboudumc to improve patient-centered care in a number of different medical specialties, with ParkinsonNet being an example [7]. The technical maintenance costs are € 5000 per annum.

In our MySAH community, patients logged in to the site using a personal digital identification code. Attention was drawn to new messages by pop-ups in a patient's mailbox. Two physician assistants and a nurse practitioner were responsible for daily communication with the community. Weekly checks on the Web-blogs were made by two neurosurgeons. General questions without the need for the intervention of a neurosurgeon were answered by the physician assistants or nurse practitioner. If the answers to questions required more specialist knowledge, responses were provided by one of the two neurosurgeons.

We used a questionnaire, which was sent to consecutive aSAH patients, to evaluate the usability and utility of MySAH. If patients were unable to complete the questionnaire, their caregivers were asked to do it for them. Those who did not return a completed questionnaire within 3 weeks were contacted by telephone and asked why. An eventual telephone evaluation was conducted by a physician assistant who was not involved in the treatment of the patients.

All of the aSAH patients referred to Radboudumc between November 1, 2012, and September 30, 2013, were candidates for participation, and all survivors were invited to take part in the research. The demographics of all of the referred patients were registered, as were the modified Rankin scale (mRS) at discharge and the type of post-hospital care (home, rehabilitation, or nursing home). The mRS is frequently used in aSAH patients to score outcomes and is an ordinal scale varying from 0 to 6 (0=No symptoms; 1=No significant disability. Able to carry out all usual activities, despite some symptoms; 2=Slight disability. Able to look after own affairs without assistance but unable to carry out all previous activities; 3=Moderate disability. Requires some help but able to walk unassisted; 4=Moderately severe disability. Unable to attend to own bodily needs without assistance and unable to walk unassisted; 5=Severe disability. Requires constant nursing care and attention, bedridden, and incontinent; and 6=Dead). The mRS scores have been dichotomized as ≤3 and ≥4 because it was assumed that patients with a score of more than 3 would use the Internet less often. Approval for the study was obtained from the local medical ethics committee (CMO Arnhem-Nijmegen).

Figure 1. Poster of access page.



Questionnaire

We developed a questionnaire that had two parts. The first of these contained general questions on perceived care, while the second asked questions on the usability and usefulness of the MySAH community. The questions were adapted from a previously published patient agreement questionnaire containing usability- related and usefulness-related statements and were expanded for use with MySAH [9]. Answers were given using a 5-point Likert scale, the data were summarized by a median, and for the analysis, the results were collapsed in two categories, with the neutral score counted on the negative side (agree/disagree). The results are presented graphically with median and interquartile ranges [10]. There was one open-ended question about what was missing from the MySAH tool. If possible, the responses were classified according to the three components of the community and with respect to suggested technical alterations.

Results

Included Patients

In total, 66 patients with aneurysmal subarachnoid hemorrhage were informed about the online health community. Two patients died in the post-discharge period. Of the 64 remaining potential MySAH users, 38 did not log in, 4 could not be contacted in the post-operative period, 3 were willing to log in after a rehabilitation period, 2 did not log in because of their clinical condition, 5 had technical difficulties logging in, 5 did not have a computer, and 19 did not provide a reason for their non-participation. Finally, 26 patients did gain access to MySAH, 20 of whom were willing to participate in the study (Figure 2). The demographics of the patients, stratified by their participation, are shown in Table 1.

The participants who evaluated MySAH were not significantly different in terms of their gender or discharge location (P=.33). However, those who did participate were younger (P=.03) and were in a better clinical condition (mRS) at discharge (P=.03).

Table 1. Patient characteristics.

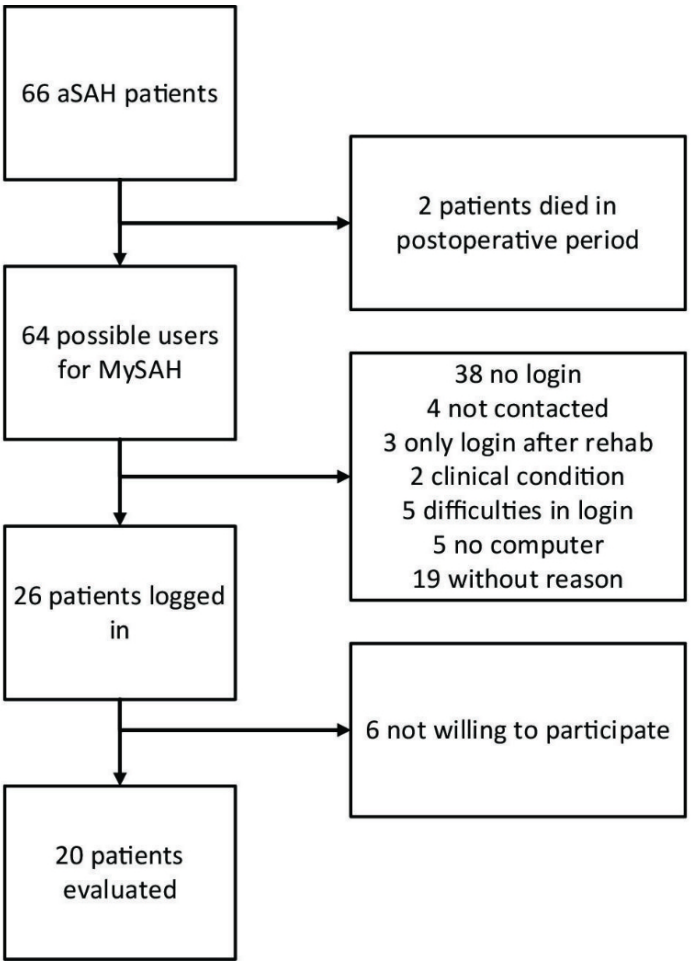
Characteristics	Participant	Non-participant	Total	*P* value for calculated difference between groups[a]
Number of patients, n (%)	20 (30)	46 (70)	66 (100)	Not applicable
Male/female, n/n	6/14	17/29	23/43	.78[b]
Age in years, median (SD)	48.5 (11.7)	56.0 (11.9)	54.0 (12.2)	.03[c]
mRS at discharge, n				.03[b]
≤3	19	31	50	
≥4	1	15	16	
Discharge, n				.33[b]
Home	14	22	36	
Rehabilitation	5	18	23	
Nursing home	1	6	7	

[a]Values are considered to be significant if $P<.05$.

[b]Fisher's Exact test (2-sided).

[c]Mann-Whitney U test (2-tailed).

Figure 2. Flow chart of patients included in study.



Patient Satisfaction and the Use and Usability of MySAH

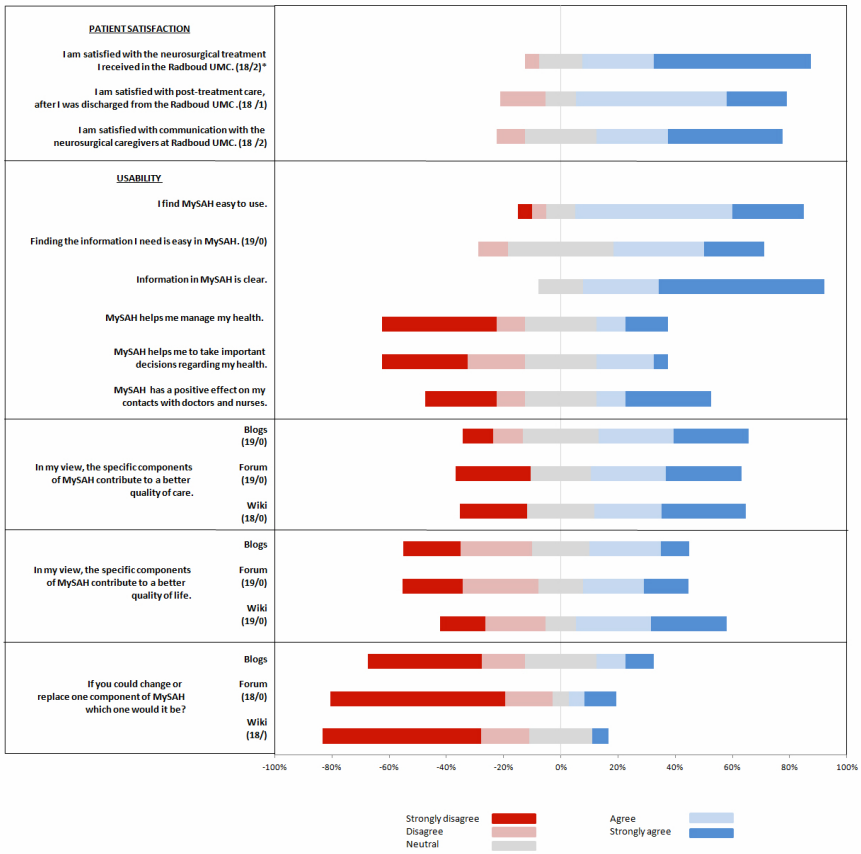
The MySAH community was used for a mean period of 7.2 months, mainly bi-monthly (9/20, 45%) or monthly (7/20, 35%). A minority used the tool weekly (3/20, 15%) or daily (1/20, 5%). In most cases (16/20, 80%), the patient was the main user of MySAH, while the other responders were proxies. No specific part of the MySAH community was used preferentially by either the patients (wiki: 4/20, 20%; forum: 10/20, 50%; blogs: 2/20, 10%; not answered: 2/20, 10%) or their proxies (wiki: 3/20,

15%; forum: 2/20, 10%; blogs: 2/20, 10%; and not answered: 12/20, 65%).

The questionnaires were mainly completed by the patients and in a minority of cases by their caregivers. Patient satisfaction with treatment, post-treatment care, and communication with caregivers was generally rated positively (see Figure 3).

The information was easy to use (4.0) and find (4.0) and was also clear (5.0). However, it was not beneficial for managing health, making important decisions regarding health (2.5), or making contact with caregivers (3.0). Patients were positive about MySAH's contribution to the quality of their care, but not to their quality of life. No specific component (blog, forum, or wiki) was preferentially rated, nor did the patients discard one aspect in particular. Most patients (18/20, 90%) would recommend the community to others in their position.

Figure 3. Patient satisfaction and usability (results of the questionnaire are depicted as a set of diverging stacked bar charts. Each stacked bar is 100% wide and partitioned by the percent of that group who have selected the agreement level indicated in the legend below the body of the plot. The legend is ordered by the values of the labels. Asterisk=answer by number of patients/caregivers).



Open Remarks

In total, 16 patients made 21 suggestions for future improvements to the community (Table 2). These responses were classified according to the three components of the community and with respect to suggested technical alterations. More frequent blogs, including by a rehabilitation specialist, was one suggestion. The forum could apparently also benefit from use by a larger number of patients overall and by patients with more positive disease experiences. The wiki section should contain more information about aftercare, psychological consequences when at home, pregnancy after aSAH, current news, and more general factors. Other suggestions were related to login and layout and navigation on the site.

Table 2. Items for improvement[a].

Item	Suggestions
Blogs	More blogs (1), in combination with a rehabilitation specialist (2).
Forum	More patient contact (1), also positive experiences (1).
Wiki / information	More information on aftercare (2), psychological consequences at home (3), pregnancy after aSAH (1), current news (2), and general information (3).
Technical	Login (1), navigation (1), layout (3).

[a]Numbers in parentheses = number of patients who suggested this improvement.

Discussion

Principal Findings

The conceptual framework of the online community, MySAH, is to improve patient care and obtain better clinical outcomes through optimizing engagement of the patient with the treatment. This is accomplished by an exchange of information between patient and caregiver and vice versa. Such a concept is comparable with sociological studies in other fields [11].

This online health community has promising features. Although the number of responses to the questionnaire was not high (30% responders), the majority graded the items concerning usability as good. The response rate is probably related to the clinical outcome after aSAH; the patients using the community were generally in better health, which means that it may not be valuable for those in a worse condition. The users of the community were also younger, which is generally the case with health-related Internet use [12].

At our center, the treatment of aSAH patients is carried out by a subspecialist team working in a multidisciplinary setting. This team consists of neurologists, neurosurgeons, neuroradiologists, neurorehabiliation specialists, neurointensivists, and a dedicated nursing team using a protocolized aftercare program. This probably contributes to patient satisfaction with treatment, post-treatment care, and communication with caregivers. As important decisions are already taken within this framework, it is likely that no additional benefit of the online health community was identified with respect to managing health, making decisions regarding health, or making contact with caregivers. Moreover, a recent study investigating the use of an online forum identified a participant's motivation to seek out information as one of the factors related to participation in an online community [13].

However, the patients were positive about MySAH's contribution to their quality of care. Indeed, with the increasing centralization of subspecialized care, this online health community can provide additional, easy access (after-) care at a distance without the need to travel [14]. This was also emphasized in the open suggestions made by the patients concerning how to improve communication with the specialists (rehabilitation specialist) involved with health care after aSAH, and could be valuable in a future online health community. Such a tool would enable answers to be provided quickly on apparently less important, but for the patient at their stage of rehabilitation, very relevant issues (eg, washing hair, biking, sex). MySAH might also serve as a tool for self-management whereby patients are helped to gain control over their lives [7]. Additionally by implementing and evaluating this online community, patient engagement has led to advancements in the aftercare, especially by improved and tailored information. A lesson learned: for future caregivers starting a community, careful selection of the possible participants and their needs is paramount.

As indicated in other publications, household Internet access in the Netherlands is about 92% and should therefore be a minor limitation with respect to access to an online health tool [7,15]. Indeed, this is in line with our data in which only five of 66 patients (7.6%) did not have a computer. However, in some other states in the European Union, Internet access is less, down to 45%, and might therefore be a restricting factor in the success of such an online community [7,15].

Health-related quality of life is significantly reduced in patients with aneurysmal subarachnoid hemorrhage [2,3,16]. Important factors associated with this are physical health issues, depression, cognitive impairment, anxiety, and fatigue [2,3,17-19], and standard aftercare and rehabilitation focuses on these problems. These impairments may, however, have been barriers to the use of the community by those who might potentially benefit from it. Those who did participate evaluated the tool neutrally, regarding their quality of life as neutral.

Limitations

This research has several limitations. First, the number of patients evaluated was only 20, as some of those approached were unwilling or unable to use the MySAH tool. However, within this pilot study, this outcome highlighted the limitations of the community in this patient category. Moreover, for evaluation purposes, having 20 participants is considered to be adequate [9]. Second, usability was self-reported, although from a quality of life perspective the use of subjective experiences is important [17]. Third, the online health community was used as an additional aftercare program and might have experienced some redundancy.

Future studies should assess the value of this online health community when fully integrated in, and as an adjunct to, face-to-face interactions. This could tailor aftercare to the wishes of the patient, enabling more patient-centered care. In our view, it should be emphasized that face-to-face contact continues to be essential in order to precisely determine outcomes and identify possible neurological deficits. Moreover, we envisage a broader use for the MySAH community in other centers involved in aSAH care in the Netherlands. Certainly, the general sections of the site could productively be used by patients from other centers, and the experiences of other caregivers would probably also be beneficial. Furthermore, a larger group of active members may possibly facilitate community sustainability [20,21]. Indeed, organizational commitment and financial and human resources are essential to maintain a community, and these efforts can be supported by the involvement of a larger group of people who provide care to aSAH patients [20]. As a result of the responses to the open questionnaire used in this pilot study, information will be added to the wiki section and rehabilitation specialists will become engaged in the MySAH community.

Conclusions

In this pilot study, the online health community MySAH contributed to the aftercare of patients suffering from aSAH. There was easy access to information that was relevant for patients and families, which could be obtained from peers or caregivers. The MySAH community will, however, mainly be useful for a select group of patients because of differences in clinical outcomes.

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Conflicts of Interest

None declared.

Multimedia Appendix 1

English translation of frequently asked questions in category “Can I…?” (In Dutch; Mag ik…?).

[PDF File (Adobe PDF File), 38KB - resprot_v3i4e60_app1.pdf]

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Abbreviations

aSAH: aneurysmal subarachnoid hemorrhage
mRS: modified Rankin Scale

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Original Paper

Examining the Use of an Open Digital Health Library for Professionals

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Abstract

Background: The Norwegian Electronic Health Library (The Library) is a website for health personnel. Most of the content is also open to the public. Usage statistics have risen sharply in the years 2010-2013.

Objective: We wanted to find out whether the rise was caused by health personnel, the general public, or other factors.

Methods: Since we lacked direct information, we had to use proxy data to shed light on our questions. We applied mixed methods (database of registered users, user survey, usage statistics, and statistics from suppliers), and triangulated between them.

Results: Health personnel were our largest user group, but The Library was also accessed by students, patients, and other groups. Content in Norwegian was preferred to English language content. Concise, practical information was preferred to more comprehensive information. Patient leaflets were the most popular information type. Mobile phone visits differed from personal computer visits both in terms of time of day and what kind of information was viewed.

Conclusions: The Library was used mostly by health personnel, as intended, but our data are inconclusive regarding a possible change in user groups. There was a large degree of consistency in results when using different investigation methods. The survey points toward health personnel being the largest user group, and the usage statistics show that patient leaflets are the most popular content, being viewed by both health personnel and patients.

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KEYWORDS

libraries, medical; access to information; information dissemination; search engine /statistics; Web log analysis

Introduction

The Norwegian Electronic Health Library (The Library) is a publicly funded website (Helsebiblioteket.no) for professionals, established in 2006. It is marketed to health personnel, but not to the general public. The Library provides free access to important sources of knowledge intended for health personnel, including point-of-care tools (reference works), bibliographic databases, and a large number of scientific journals [1].

The Library is also a sharing platform for guidelines, patient leaflets, and clinical procedures. It contains links to all its purchased sources, as well as open sources like rating scales, reports, summarized research, patient leaflets, and guidelines published elsewhere.

Thus, The Library is partly a traditional library service with purchased content, and partly a sharing platform where information resources are published.

Most of the content is available to anyone with a Norwegian Internet protocol (IP) address, including free access for the entire Norwegian population [2] to the five largest general medical journals and point-of-care tools like “UpToDate” and “BMJ Best Practice”. To our knowledge, The Library is the only one

with national licenses for major international journals and point-of-care tools.

Users do not have to be logged in when using open access sources or subscribed material with a national access license, and may go directly to the original content provider, such as a journal's home page. Some of the content is, for economic reasons, only available to health personnel and students. Such access is given according to recognized institutional IP addresses (hospitals, health institutions, universities, university colleges) or personal username and password assigned from The Library. As long as users are at their workplace, the IP address is recognized, so they don't have to log in. If users are at home or anywhere else outside of the workplace they must log in to get full access to all of The Library's resources.

From 2010 to 2013, The Library saw a sharp rise in usage. We investigated the use of The Library to find out whether this rise was caused by professionals, the general public, or other factors.

Methods

Overview

Since information on the individual visitors to The Library is not recorded, we had to rely on proxy variables. A proxy variable is something that is probably not in itself of great interest, but from which a variable of interest can be obtained [3].

Proxy variables serving as indicators of who is using The Library could be: where users are coming from, at what time of day the website is visited, what kind of information is most frequently used, and what kind of device people use. We collected data from several sources in order to shed light on the issue from different angles.

The data were taken from The Library's database of registered personal users, a survey of our users within a given period, usage statistics for our website, and usage statistics from our content suppliers.

Database of Registered Personal Users

The Library has a database of registered personal users, but we do not record their activities on the website. We have information on professional background for 48,950 of 85,270 registered users (data extracted March 31, 2014), but the quality of the data varies.

Survey

We did a user survey from October 1 - October 9, 2013, asking the website visitors if they were health personnel, and if so, which professional group they belonged to.

The users who came to the website were served a pop-up that they could accept or reject to take part in the survey. The survey pop-up was not displayed again after the users had accepted or rejected it.

We asked the participants whether they were health personnel, students, patients/dependents, non-health personnel employees in the health services, researchers, or "other". If they were health personnel, they were asked which personnel group they belonged to. Health personnel were also asked which sector they worked in.

Usage Statistics

We analyzed usage statistics for The Library website from the years 2010-2013 by Google Analytics, and we looked particularly at "pageviews" of certain types of information like patient leaflets and guidelines. A "pageview" is recorded each time a user visits a webpage [4]. A single visitor can conduct many pageviews on a website. Each time the visitor returns to the page, a new pageview is recorded. Google Analytics distinguishes between new visitors and returning visitors by using cookies [5].

We analyzed bounce rates for different parts of our website. Bounce rate is defined by Google as the percentage of single-page sessions, that is, sessions in which the visitor left the site from the entrance page without interacting with the page [6].

Sessions are defined by Google as the number of individual sessions initiated by all the users of a site. If a user is inactive on the site for 30 minutes or more, any future activity is attributed to a new session. Users who leave the site and return within 30 minutes are counted as part of the original session [7].

In the analysis of pageviews for guidelines versus patient leaflets, we had data for only the first five months of 2014; therefore, we extrapolated the data as if the numbers were representative for the whole year.

Statistics From Content Suppliers

We used available statistics from our suppliers of bibliographic databases, journals, and point-of-care tools to get data on the usage of their resources. The statistics for journals are based on successful full text downloads according to the COUNTER standard (Counting Online Usage of Networked Electronic Resources) [8]. There is a slight difference between this and a pageview, but for the practical purpose of our comparison we have chosen to ignore this. The statistics on bibliographic databases are based on number of executed searches (number of times a user has pressed the Search button) according to the COUNTER standard. The usage statistics cover the period 2010-2013, and the sources for these statistics are shown in Table 1.

The Norwegian point-of-care tool Handbook of Emergency Medicine (Legevakthåndboken) became available in September 2012. Our statistic for the Handbook of Emergency Medicine is based on Google Analytics.

In our comparison of the statistics from the suppliers with the usage statistics for our own website, we have regarded a full text download or a search as a pageview. We did this to be able to compare the data from different sources.

Table 1. Suppliers.

Bibliographic databases	Journals	Point-of-care-tools
OVID (AMED, Embase, Medline, Ovid Nursing, PsycINFO)	Ovid (LWW, Nursing Full Text)	UpToDate
Ebsco (Cinahl)	Informa Healthcare	BMJ (Best Practice)
	APA (PsycARTICLES)	Pharmaceutical Press (BNF Children og Martindale)
	ProQuest	Lexicomp
	BMJ (23 titles)	Gyldendal Akademiske (Handbook of Emergency Medicine, in Norwegian)
	JAMA Network (10 titles)	
	The Lancet (4 titles)	
	Annals of Internal Medicine (including ACP Journal Club)	
	New England Journal of Medicine	

Results

Database of Registered Users

In the database of registered personal users, we have information on professions for 56.98% (48,590/85,270) of the users (data extracted March 31, 2014). The largest group of personal users was nurses, followed by physicians, physiotherapists, psychologists, pharmacists, and assistant nurses (see Table 2).

Table 2. Profession and number of registered personal users of The Library (Helsebiblioteket.no).

Profession	Number of registered users n=48,590 n (%)
Nurses	12,166 (25.04)
Physicians	8924 (18.37)
Physiotherapists	2503 (5.15)
Psychologists	1806 (3.72)
Pharmacists	1070 (2.20)
Assistant nurses	804 (1.65)
Engineers	219 (0.45)

Survey

All in all, 2563 (4.27%) visitors took part in the 9-day survey out of an estimated number of approximately 60,000 unique visitors over the same period. Not all respondents answered all the questions of the questionnaire.

A total of 55.95% (1434/2563) of the respondents reported to be health personnel, 15.02% (385/2563) students, 11.86% (304/2563) patients/dependents, 5.93% (152/2563) employees of the health services (not health personnel), 4.37% (112/2563) researchers, and 6.87% (176/2563) other.

Among the health personnel (n=1438), physicians were the largest group (38.66%, 556/1438), followed by nurses (29.62%, 426/1438), psychologists (8.34%, 120/1438), physiotherapists (5.63%, 81/1438), and pharmacists (4.66%, 67/1438).

The respondents were asked which sector of the health services they worked in: 65.51% (940/1435) of health personnel came from hospitals and specialist health services, 28.08% (403/1435) from primary care, and the rest from educational institutions, industry, research, etc.

Usage Statistics

From our usage statistics, we can see that patient leaflets had the sharpest increase in pageviews from 2011 through 2013, and it was the most frequently viewed information type in 2013 (see Figure 1). There was a sharp increase in pageviews of guidelines in the period 2010-2013.

The Handbook of Emergency Medicine immediately became very popular and this accounts for a large proportion of the growth in pageviews for point-of-care tools in 2012 and 2013 (see Figure 2).

The share of users coming from Google and other search engines increased throughout the period 2010-2013 and constituted approximately two-thirds of the traffic in 2013. The share of new visitors increased steadily from 2010 through 2013.

Each visitor spent a shorter time per session on the website in 2013 than in 2010.

The share of visitors using mobile phones increased from 1.00% (10,074/1,007,395) in 2010 to 21.00% (552,859/2,632,660) in 2013. This is a general trend for websites and The Library does not differ considerably from other websites in this respect. Mobile phone users stayed for a shorter length of time on the site than personal computer (PC) users. They also viewed fewer pages than PC users. Mobile phone users spent much more time on patient leaflet pages than on guideline pages. Mobile phone users also viewed more patient leaflet pages than guideline pages. Pageviews of patient leaflets increased throughout the day, while pageviews of guidelines peaked during office hours (see Figure 3).

While visits by PC users peaked in the office hours, visits by mobile and tablet users increased steadily throughout the day (see Figure 3).

More users came from recognized workplace networks in 2013 than in 2010, but the relative share of these decreased (see Table 3).

From 2010 to 2013, we saw a 2% increase in nightly use (midnight to 8am) and a 4% increase in evening use (4pm to midnight), and a relative reduction in office hours (8am to 4pm) use.

People who viewed patient leaflets spent 4 minutes or more on each page and the bounce rate was very high.

Based on the usage statistics, the most frequently used type of information from the website was patient leaflets (see Figure 1). Other frequently used types of information on the website were guidelines and procedures.

Both the number of documents (patient leaflets, guidelines, and procedures) as well as the number of topics covered by these documents increased sharply from 2010 to 2013.

We found a difference between the kind of information that was viewed by mobile users and PC users. Patient leaflets were viewed on mobile phones almost as frequently as on PC screens. Guidelines were much more frequently viewed on PCs (see Figure 4) and very rarely on mobile devices.

Table 3. Usage statistics for The Library website (Helsebiblioteket.no).

Usage	2010	2011	2012	2013
Number of pageviews - all days	3,361,563	3,416,930	4,309,538	5,737,733
Number of pageviews - weekdays	2,852,482	2,903,624	3,604,040	4,724,635
Number of visits	1,007,395	1,125,686	1,605,288	2,632,660
Search engine traffic visits, n (%)	312,292 (31.00%)	427,761 (38.00%)	931,067 (58.00%)	1,676,394 (63.68%)
Direct traffic visits, n (%)	392,884 (39.00%)	382,733 (34.00%)	353,163 (22.00%)	579,185 (22.00%)
Other traffic visits, n (%)	302,219 (30.00%)	315,192 (28.00%)	321,058 (20.00%)	368,572 (14.00%)
Bounce rate	49%	55%	63%	72%
Pages per visit	3.34	3.04	2.69	2.18
Time spent per visit	4m15s	3m25s	2m38s	1m56s
New visitors	366,786 (36.41%)	453,252 (40.26%)	771,366 (48.05%)	1,308,465 (49.70%)
Mobile phones, % of visits	10,074 (1.00%)	33,771 (3.00%)	144,476 (9.00%)	552,859 (21.00%)
Tablets, % of visits	0%	0%	6%	13%
Time spent per visit, desktop	4m17s	3m36s	3m3s	2m25s
Time spent per visit, mobile phone	2m14s	2m21s	40s	40s
Pages per visit, desktop	3.33	3.05	2.90	2.61
Pages per visit, mobile phone	2.43	2.62	1.39	1.24
Visits from “workplace” networks	511,845	546,407	654,617	903,048
Office hours[b] pageviews, % of total on weekdays	1,965,360 (68.90%)	1,991,886 (68.60%)	2,378,666 (66.00%)	2,962,346 (62.70%)
Evening hours[c] pageviews, % of total on weekdays	761,613 (26.70%)	772,364 (26.60%)	1,027,151 (28.50%)	1,450,463 (30.70%)
Overnight hours[a] pageviews, % of total on weekdays	125,509 (4.40%)	142,278 (4.90%)	198,222 (5.50%)	311,826 (6.60%)

[a]overnight hours: midnight to 8am

[b]evening hours: 4pm to midnight

[c]office hours: 8am to 4pm

Figure 1. Pageviews of different information types on The Library website, Helsebiblioteket.no.

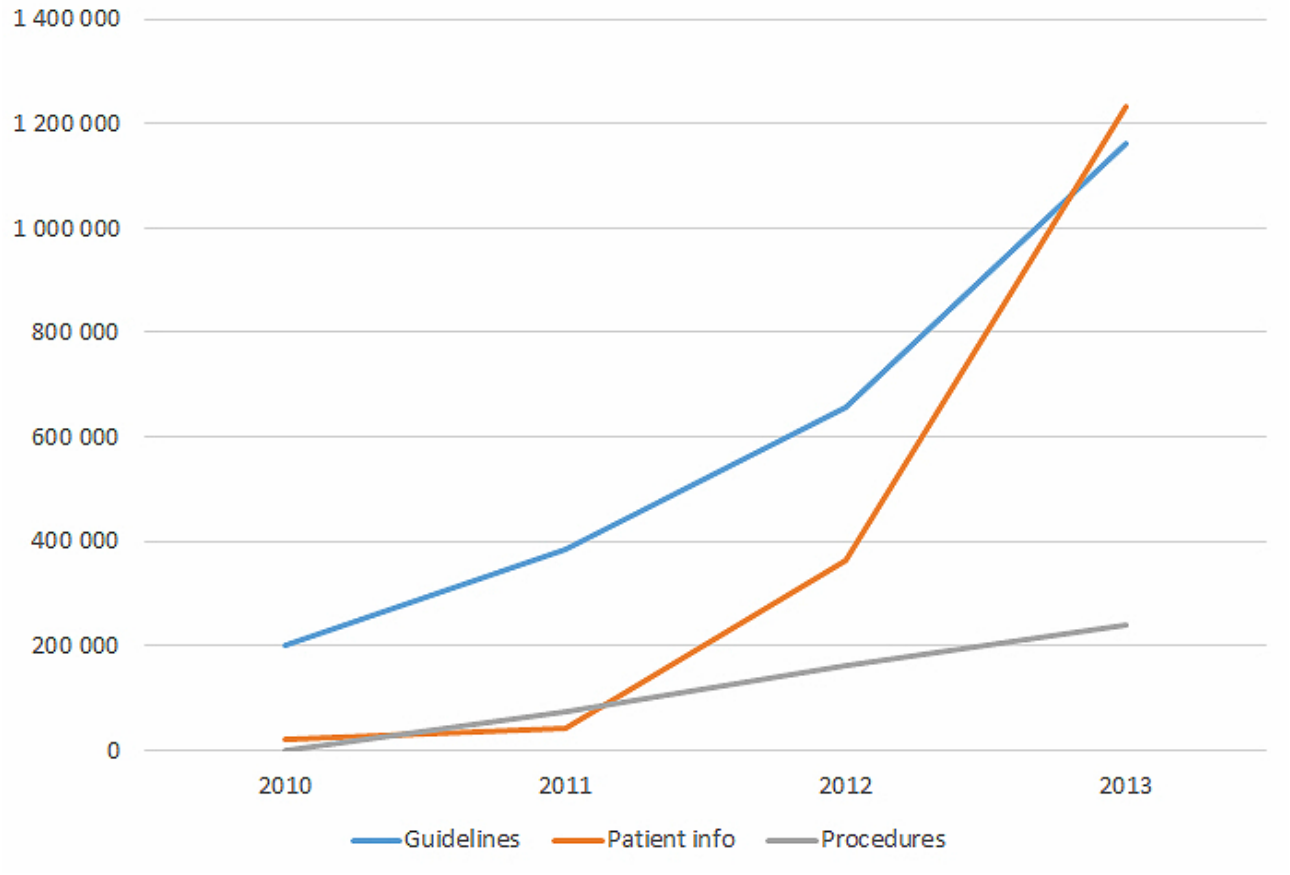


Figure 2. Pageviews of The Library website (Helsebiblioteket.no) versus purchased content.

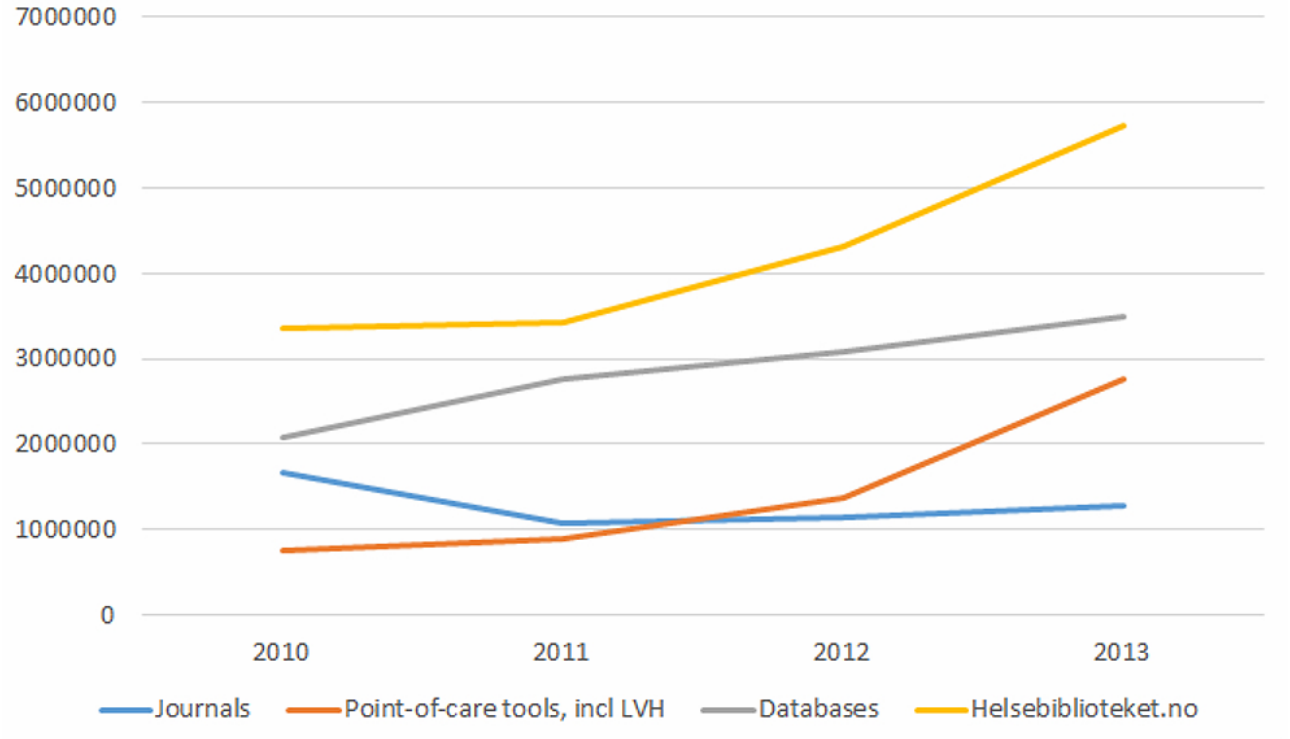


Figure 3. The Library (Helsebiblioteket.no) pageviews and time of day of patient information leaflets (PI) in 2013.

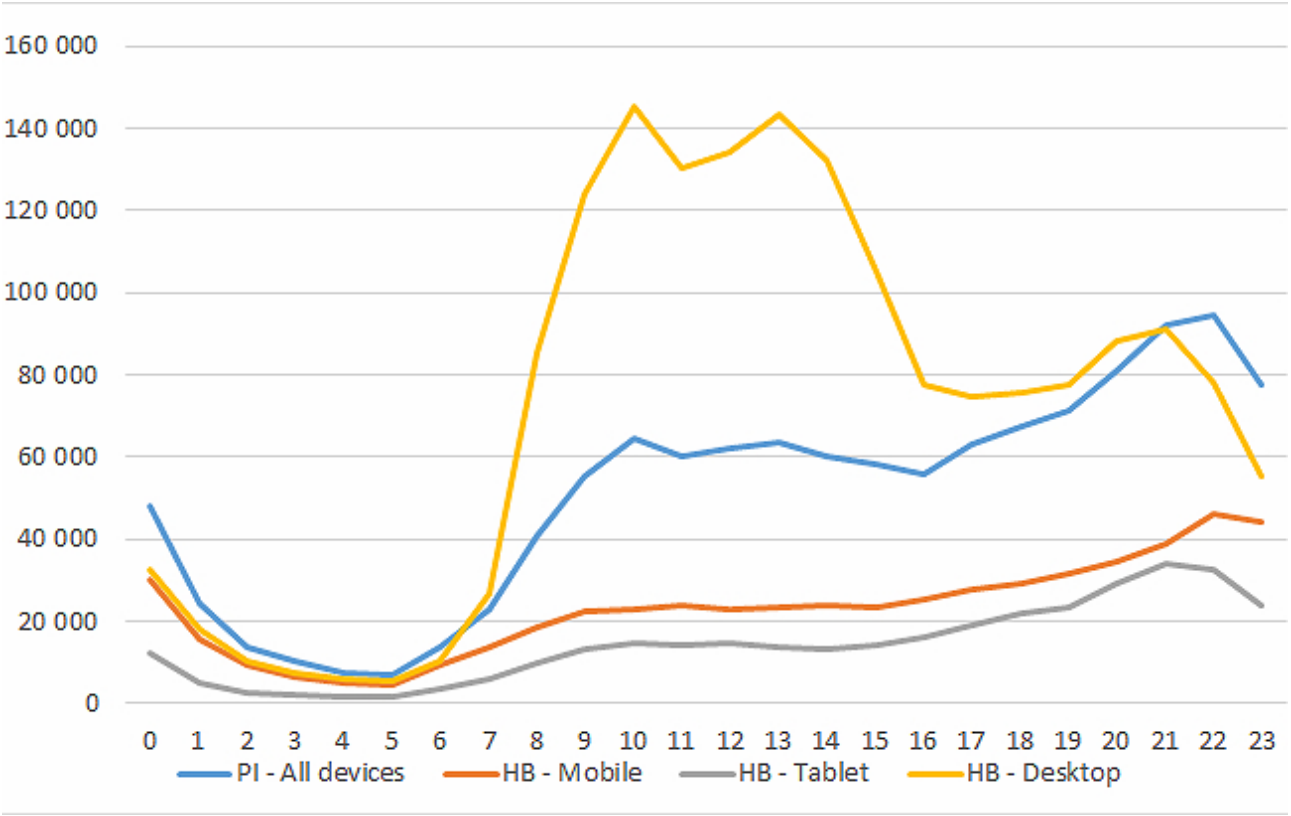
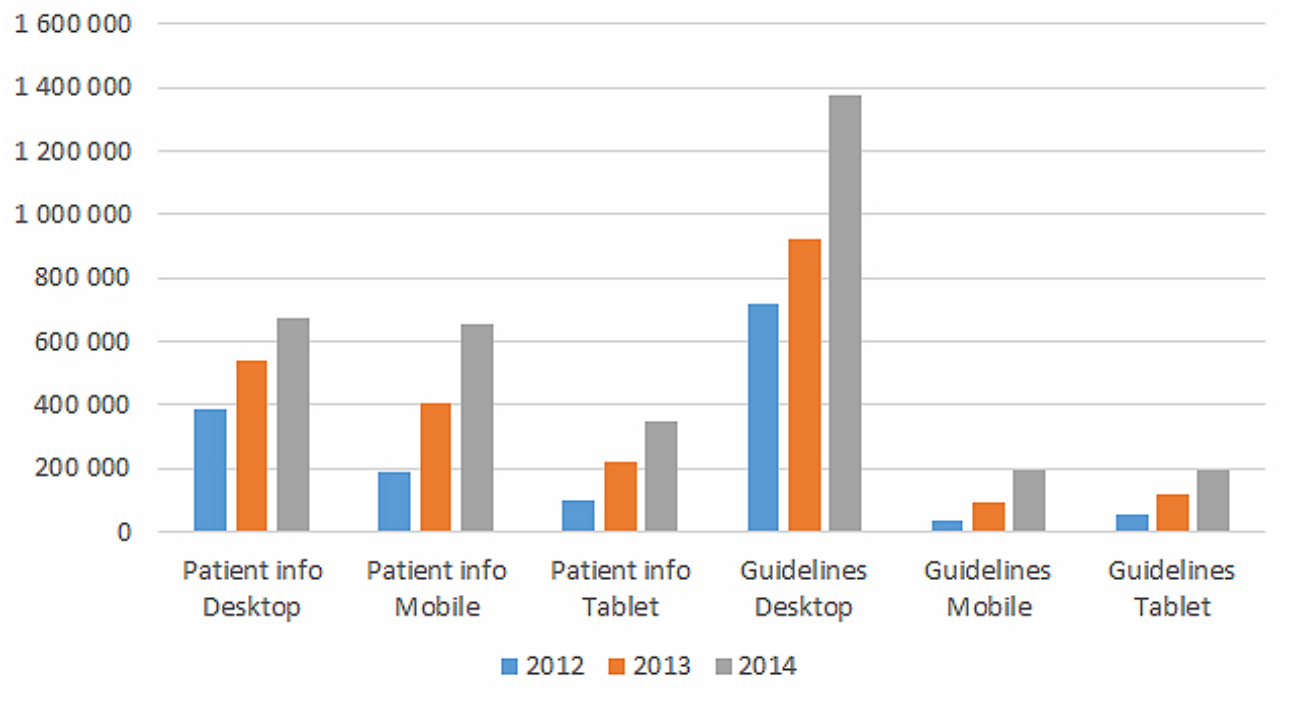


Figure 4. Pageviews of patient information leaflets and guidelines 2012-2014. Estimates for 2014 are based on the months January to May.



Statistics From Content Suppliers

Based on statistics from our suppliers (Table 1), the usage of journals, point-of-care tools, and bibliographic databases showed a steady growth in the period we analyzed (2010-2013).

The statistics from our suppliers combined with the usage statistics (see Figure 2) make it possible to compare pageviews of journals, point-of-care tools, database searches, and The Library website. Pageviews in the website increased more than the pageviews of purchased sources. In 2013, there were 5.7 million pageviews of the website pages, against 4.0 million pageviews for purchased resources like point-of-care tools and journals. Point-of-care tools rose sharply from September 2012 when the Norwegian language point-of-care tool Handbook of

Emergency Medicine was introduced. The Handbook of Emergency Medicine is available online to anyone with a Norwegian IP. It can also be downloaded as an app. This source now has more pageviews than all other point-of-care tools combined.

Discussion

Principal Results

The Library website and its purchased content are widely used by both health personnel and the general public. Over the last few years, there has been a sharper rise in the use of the website than in the use of most content located outside the website. This could be due to better visibility in Google and other search engines, but that is probably not the only reason. There has also been an increase in the number of documents published on the website.

Norwegian language seems to play a role. Norwegian guidelines, patient leaflets, and the Handbook of Emergency Medicine had a sharper rise in popularity than the rest of our content. In fact, the Handbook of Emergency Medicine had more pageviews than all the other point-of-care tools combined in 2013. A study among Norwegian physicians has shown that it is highly valued to get information in their mother tongue [9].

Cultural factors, ease-of-use, and how practical the information is, may also play a role. The Handbook of Emergency Medicine is very concise and practical, but it covers only a fraction of the topics that our English language point-of-care tools cover. Patient leaflets are also very concise and highly popular among our users. A study on preferred sources of information for primary care physicians from 1997 states that “Information resources for answering clinical questions should be readily available, familiar, and quick to use” [10].

There was large growth in the number of mobile phone users visiting the website. We found that visits from PC users peaked during office hours, while visits from mobile users increased throughout the day. Nicholas et al [11] and Cronk [12] have shown that there is a difference in the time of day when PC users visit a website and when mobile users visit the same site, and the difference is the same as we found: PCs peak in office hours and mobile phones peak in the evening. Even so, we still have more PC users in the evening than we have mobile users.

Nicholas et al [11] found that mobile users typically viewed other kinds of material than PC users, typically shorter texts and fewer pages. We found the same—mobile phone users viewed relatively more patient leaflets and fewer guidelines than PC users on The Library website.

Strengths and Limitations

The use of different methods to analyze the usage is a strength of this study. It is also a strength that we have followed the usage over several years.

There are some weaknesses regarding the database, survey, and the Google Analytics data.

The data quality of the database of registered personal users is not optimal. The users report their professional position in free text, which makes classification difficult. Our survey covered only 9 days, and the response rate was low. According to the survey software supplier Surveygizmo [13], external surveys like customer satisfaction surveys generally get 10-15% response rates, while internal surveys get 30-40% response rates. Our survey had an estimated response rate of 4%. This estimate is probably too low, since we didn’t measure non-responders directly, but based the estimate on Google Analytics’ unique visitor statistic and cookies.

Google Analytics is based on samples of data and hence may be skewed. We have, however, followed the use of the website through Google Analytics over 4 years, and the tendencies are consistent over all these years.

Google Analytics’ new/returning visitor statistic may be unreliable since it uses cookies. Cookies are specific for each Web browser and device, so the same person will be counted as a new visitor if he or she first visits a site from the office PC, then from the home PC, and again if he or she changes Web browser, deletes cookies, or visits the same site from a mobile phone. The reported ratio of new users is therefore probably exaggerated [5]. The same applies to statistics of unique visitors.

Some of the Google Analytics data give a skewed picture even if they are technically correct. Guidelines pageviews, for example, are reported a bit too high, due to each guideline being split into several documents on our website.

Interpretation

The data from the usage statistics for the website indicate that patient leaflets are becoming more popular. At the same time, the survey data indicate that the majority of users are health personnel. How can this be explained?

The Library is primarily a link portal. Patient leaflets are, along with guidelines, recommendations, and procedures, the only content that is published on the website itself. The same development as we have seen for patient leaflets can be seen for guidelines. Their use (registered as pageviews) also increases sharply. In other words, what we are seeing is an increase in the usage of content that is located on the website itself.

The percentage of users coming from the Google search engine underpins this. The content of the website is more visible to Google than the content of our purchased material. The proportion of users coming from search engines has increased from 31% to 64%.

The proportion of new visitors increased from 36% to 50% over three years. If this is real change and not just an artefact due to the use of cookies, this could very well be a sign that the search engine optimization is starting to pay off. The Library website, Helsebiblioteket, has a Google PageRank of 7/10, which is regarded as very high (on par with the newspaper The Guardian). Google PageRank is an algorithm used by Google Search to rank websites in their search engine results. PageRank is a way of measuring the importance of website pages [14] and is given as a number between 0 and 10 [15].

The high Google ranking may lead more people from outside the ranks of health personnel to our pages. This may be one of the reasons why time spent on the portal and pages per visit

both go down. Some of these visitors probably don't find what they expect on the website.

Is it patients then who read the patient leaflets? Our data give us an indication, but are not conclusive.

The hour of the day of viewing of patient leaflets shows a pattern that resembles both the visits by PC users and the visits by mobile phone users. There is a small peak in office hours, but the number of pageviews increases again from 4 pm and peaks at 10 pm. This indicates that a large proportion of the readers of patient leaflets are the general public or patients, or at least not people at work. But the peak in office hours indicates that the patient leaflets are also used by health personnel at work.

We have to remember that most health personnel are not physicians, and they might very well make good use of patient leaflets. It may also be that physicians visit the website and print the patient leaflets for their patients. Professionals and patients alike seem to embrace simple, short, and trustworthy information.

The inclusion of the patient in the decision process is becoming more common in health care. To make this inclusion meaningful, the patients need to be informed. According to Longo [16], patients have different information needs depending on where they are in the process. Since The Library is quite unique with its national licenses for highly respected point-of-care tools and journals, we didn't find any other studies examining patients' use of similar websites.

The importance of visibility in search engines for medical websites was shown by Giustini as early as 2005 [17]. This visibility also makes it possible to retrieve professional information using devices like mobile phones far away from the office.

We found two studies on how a large public website is accessed with different devices. Cronk studied the use of the British government website gov.uk and found that mobile phones and tablets were used more in the evenings and weekends, while desktop/laptop PCs dominated the office hours [12]. Nicholas et al compared mobile users with PC users of Europeana, the European fulltext cultural website. They found that mobile users were the fastest growing group and that their visits were different from PC user visits. Mobile visits were typically shorter, less interactive, and less content was viewed per visit. Mobile use peaked in the evenings and weekends. They found that tablet users behaved very much like PC users [11].

We found the same pattern as Cronk [12] and Nicholas et al [11]. PCs peaked during working hours, while mobile phones increased throughout the day and peaked around 10 pm. We also found that pageviews of patient leaflets followed a pattern in between the PCs and mobile phones. Patient leaflets showed a small peak during office hours and a larger peak in the evening. This indicates that patient leaflets are actually read by the general public, but they are probably also read or handed out by health personnel during office hours.

We are now redesigning The Library website, partly to make it more mobile friendly, and the findings in this study are being taken into consideration in this process. It was decided to keep the patient information leaflets, partly as a consequence of the findings in this study.

Conclusion

We assume our largest user group is health personnel and students, as intended. The Library is widely used among health personnel, and to some extent also by the public. Norwegian language content is more popular than English language content. We cannot conclude whether the high and increasing popularity of patient leaflets is caused by patients using The Library more than before.

Conflicts of Interest

All authors are employees of the Norwegian Knowledge Centre for the Health Services, the parent organization of the Norwegian Electronic Health Library.

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Abbreviations

COUNTER: Counting Online Usage of Networked Electronic Resources
IP: Internet protocol
PC: personal computer

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Original Paper

Acceptability of a Web-Based and Tailored Intervention for the Self-Management of Pain After Cardiac Surgery: The Perception of Women and Men

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Abstract

Background: Approximately two thirds of adults undergoing cardiac surgery suffer from moderate to severe postoperative pain. Assisting patients with pain management is therefore critical to prevent its negative consequences. Information technologies have become part of our lifestyle and can facilitate the implementation of interventions to manage pain in a busy care setting. A computer-tailored and Web-based intervention—referred to as SOUtien à L'AutoGEstion-Traitement-Assistance Virtuelle Infirmière-Enseignement (SOULAGE-TAVIE)—for the self-management of pain was developed. Findings from a previous pilot randomized controlled trial (RCT) provided some evidence of the feasibility and preliminary effectiveness of this intervention in decreasing pain interference with a few postoperative activities and by modulating pain beliefs and analgesic intake. However, its acceptability from the patient's perspective remains unclear. Moreover, the proportion of women is much lower in the cardiac surgical population, making it difficult to detect differences in experiences between men and women.

Objective: The objectives were (1) to describe SOULAGE-TAVIE's acceptability from the perspective of adults experiencing pain after cardiac surgery and (2) to compare the perceptions of men and women.

Methods: A mixed-method approach was used to capture the various attributes of patients' perceptions of the intervention's acceptability and to compare the perceptions of men and women. Quota samples of men (n=10; mean age 62.5 years, SD 7.3) and women (n=10; mean age 64.3 years, SD 10.7) who had cardiac surgery in the past month were invited to view the intervention, complete a brief questionnaire rating its acceptability, and then to discuss each component in a 60-minute, semistructured interview. Mann-Whitney U tests were used to compare groups. The transcripts were content analyzed to generate themes based on patients' experiences with the intervention and reports of acceptability. The content of each category and subcategory were compared between men and women. Frequency counts were also done to validate the emergence of a difference between the 2 subgroups.

Results: Participants perceived the intervention to be very acceptable in terms of content and format, and tended to describe awareness-raising and convenient support experiences. Women scored higher than men in terms of the intervention's appropriateness (U=13.5, *P*=.008). They were willing to adhere to the intervention based on the importance and relevance of the advice provided, whereas men were more focused on the delivery mode and its flexibility.

Conclusions: This study underlined the acceptability of computer tailoring and persuasive communication to modulate pain beliefs and attitudes in an acute care context. Both men and women appreciated the Web-based interface and general self-guided approach of the intervention. The delivery of SOULAGE-TAVIE across the continuum of care seems to be an interesting avenue to influence the transition from acute to chronic postoperative pain.

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KEYWORDS

pain, postoperative; surgery, cardiac; patient education; Internet; mixed method

Introduction

Approximately two thirds of adults undergoing cardiac surgery suffer from moderate to severe postoperative pain and 75% of adults suffer from pain when breathing and coughing for as long as 7 days after surgery [1-3]. Several studies also showed that intensity of acute postoperative pain predicted the presence and severity of pain after discharge and is a risk factor for the development of chronic postoperative pain (CPOP) [2-5]. Indeed, pain may become chronic in 17% to 56% of adults in the 2 years following cardiac surgery, potentially compromising their recovery and daily functioning [2,3,6,7]. These prevalence rates are substantial considering that cardiac surgeries rank among the most frequent surgical procedures [8]. Therefore, assisting patients with pain management is critical to prevent its negative consequences. Individual beliefs and attitudes regarding pain and its relief interfere with the communication of pain and its management, which could partially explain inadequate levels of analgesic consumption observed in many patients following cardiac surgery [9,10]. Computer-tailored and Web-based interventions have been recognized for their efficacy on information integration and behavioral change [11-14]. Information technologies have become part of our lifestyle and can facilitate the implementation of interventions influencing pain management in a busy care setting.

SOUtien à L'AutoGEstion-Traitement-Assistance Virtuelle Infirmière-Enseignement (SOULAGE-TAVIE), which translates into self-management, support treatment, virtual nursing assistance and education, is a computer-tailored and Web-based intervention developed for the self-management of pain after cardiac surgery [15]. Its home page is shown in Figure 1. This intervention begins with a brief, 5- to 10-minute screening of the patient's pain-related beliefs and attitudes [16,17]. The second part consists of a 30-minute, tailored, preoperative session on a laptop computer facilitated by a virtual nurse that guides the participant through an animated, interactive learning process about the management of pain. The integration page for the website is shown in Figure 2. The information and strategies provided are specifically tailored to the participants' profiles of pain beliefs and attitudes and are delivered according to a predetermined algorithm and to on-screen answers. Computer tailoring was offered as a complementary and personalized intervention to empower patients without adding a burden to patients and clinicians in the accelerated context of acute care.

A pilot randomized controlled trial (RCT) was conducted to examine the feasibility and preliminary effects of SOULAGE-TAVIE [18]. A total of 60 patients were randomly assigned to the experimental group (SOULAGE-TAVIE) and the control group (usual care including an educational pamphlet). Data were collected on admission and from day 1 through 7, postsurgery. Outcomes were pain intensity, pain interference with postoperative activities, individual pain barriers, pain catastrophizing, and analgesic consumption. Findings from this previous study provided initial evidence that the intervention was feasible and helpful in decreasing pain interference with a few postoperative activities, especially breathing and coughing. The intervention also influenced the way people coped with pain in modulating some individual barriers toward pain relief and opioid analgesic consumption. A brief questionnaire also revealed participants' global satisfaction toward the intervention [15].

To our knowledge, SOULAGE-TAVIE is the first intervention of its kind in the acute care setting. Even though the feedback was positive, the results did not reveal the participants' perceptions and preferences about the content and format of the intervention, and its acceptability remains unclear. Acceptability reflects the patients' views of the intervention and can influence the implementation, patients' adherence, and consequently, clinical outcomes [19,20]. Moreover, the experience of pain after cardiac surgery has been shown to be different between men and women [21-23]. The proportion of women is much lower in this population, therefore the experiences of male patients still influence our knowledge and interventions [23]. Research on Web-based and computer-tailored interventions is quite recent and because of its consistency with a patient-centered tailoring approach [11,14,24], a description of patients' perceptions of SOULAGE-TAVIE's acceptability would help illuminate its utility and value for the practice setting [19,25]. Therefore, the objectives of this study were (1) to describe SOULAGE-TAVIE's acceptability from the perspective of adults experiencing pain after cardiac surgery and (2) to compare the perceptions of men and women.

Figure 1. Homepage of the SOULAGE-TAVIE website showing functions to determine patient profile and to start the intervention.

Figure 2. Animated integration page of the SOULAGE-TAVIE website displaying the nurse's advice on pain and anxiety, and the patient's navigation options for more information.

Methods

Design

A mixed-method approach was used that captured both quantitative and qualitative data, but the latter were more heavily weighted (quan-QUAL). This approach captured the various attributes of patients' perceptions of the intervention's acceptability and compared the perceptions of men and women. Eligible patients were invited to complete a brief quantitative questionnaire to rate the acceptability of each intervention's components before discussing each component in a semistructured interview with the interviewer.

Sample

A quota sampling strategy was used to ensure adequate representation of men and women in the study sample. It is important to note that this study was undertaken in a different health setting and with a distinct sample than the previous pilot study. Because of feasibility issues, it was not possible to

interview participants from the pilot RCT. Conducting an interview just before surgery or in the immediate postoperative phase would have been unrealistic considering the condition of the patients. The number of participants was guided by the principle of data saturation. However, 12 interviews are usually necessary to reach this point [26]. In total, 10 interviews with women and 10 interviews with men were conducted. The sample consisted of patients 21 years and older, who first had cardiac surgery involving a sternotomy—coronary artery bypass graft, valve replacement—within the past month. This time frame was selected since perceptions of the preoperative period and early postoperative pain experience would still be recent in mind, and would allow patients to participate in a 60-minute interview without causing undue fatigue. Moreover, the 6-week recovery period after cardiac surgery is recognized as challenging for patients, and pain is still very present [21,27].

Procedure

A nurse asked eligible patients at the time of the follow-up—1 week after discharge—if they were interested in participating in the study. The nurse then communicated their contact information to the principal investigator (PI, GM), who called them to explain the study and arrange an interview within 1 month. Consent forms were signed at the time of the interviews. Semistructured interviews of approximately 60 minutes were conducted. Individual interviews were favored over focus groups to increase the feasibility of the study and to avoid group dynamics that could discourage the expression of divergent perceptions among participants [28]. Interviews took place in patients' homes to avoid feasibility issues, such as coordinating the interview with the follow-up medical appointment or asking participants to travel for the interview during this recovery period. However, this choice was also methodological as location shapes interactions and relationships [29,30]. Choosing the participants' homes could mitigate the traditional relationship between patients and health professionals and allow for opinions and preferences to be voiced.

The participants started by completing the sociodemographic and postoperative pain questionnaires, including presence of pain in the last month (yes/no), intensity pattern since discharge (decrease, increase, disappearance), and frequency in the last 7 days (continuous, occasional, absent). The interview began by using the tailored and Web-based intervention. After viewing the session, the participant was requested to rate each component in terms of its acceptability. Patients' ratings of each component were used to solicit feedback on acceptability. The interviewer (GM) then invited each participant to comment on the acceptability of the intervention and on the need for, and nature of, modifying the components to fit their preferences.

Instruments

The postoperative pain questionnaire is based on the Brief Pain Inventory (BPI) [31]. The BPI includes ten items: three items focus on pain intensity (0 for "no pain" to 10 for "worst possible pain"), and seven evaluate the impact of pain on general activity, mood, walking, work, relationships, sleep, and enjoyment of life. Participants were asked to base their ratings on their pain experience in the previous 7 days. Each item represents a subscale and can be scored and analyzed individually (0-10), with the anchors being "does not interfere" (0) and "completely interferes" (10). The internal consistency was supported (Cronbach alpha between .77 and .91) [31]. Some items were added in the context of the present study to measure the pain-related impact on appetite, concentration, and breathing/coughing. This version has been successfully validated with cardiac surgery patients [9,32] and was used by the investigator in a previous pilot study [18].

The intervention components were rated in terms of four attributes: (1) appropriateness in helping patients manage pain following cardiac surgery, (2) effectiveness in promoting pain management, (3) suitability, and (4) willingness to adhere, with the use of the treatment acceptability and preference (TAP) measure [33]. The ratings refer to a 5-point scale ranging from "not at all" (0) to "very much" (4). A total scale score between 0 and 4 was obtained as a mean of the scores from four items to reflect perceived intervention acceptability. The four items demonstrated internal consistency reliability (Cronbach alpha>.80) [33]. Three subitems were added to refine the rating, and consequently, the description of effectiveness, appropriateness, and suitability. The main author of the instrument validated the content of this adaptation. Table 1 presents the definitions of each acceptability attribute of the TAP measure according to Sidani et al [19].

An interview guide was developed and reviewed by two researchers (GM, MP) familiar with intervention and qualitative research. Table 2 shows questions from the semistructured interview guide.

Table 1. Definitions of acceptability attributes of the TAP measure [19].

Acceptability attribute[a]	Definition of attribute
Effectiveness	Perception of the extent to which the intervention is helpful in the short and long terms.
Appropriateness	Perception of the intervention's overall reasonableness (ie, how logical).
Suitability	Judgment of the intervention's intrusiveness and disruption in life (how easy, how long, etc).
Adherence	Extent to which patients are willing to follow or adhere to treatment.

[a]Acceptability is defined as the patients' understanding of the treatment based on multiple elements.

Table 2. Semistructured interview guide adapted from the TAP measure [33].

Themes	Questions
Effectiveness	What do you find the most/least helpful about the computer-based program?
	In what way do you think the program would/would not have helped you manage your pain after surgery?
	In what way do you think the program would/would not have helped you decrease the impact of pain on your recovery?
Appropriateness	What do you find appropriate/not appropriate about the computer-based program?
	What strategies seem appropriate/inappropriate for managing postoperative pain?
	In what way are the strategies appropriate/not appropriate to pain management after surgery?
	What additional information (if any) would you like covered by the computer-based program?
Suitability	What pain management strategies in the computer-based program do you find suitable/not suitable?
	What do you think of the timing of the intervention?
	What do you think of the length of the intervention?
	What do you think of the virtual nurse?
Willingness to adhere	What is easy/not easy about using the computer-based program?
	What is easy/not easy about applying all the strategies?
	What (if anything) could be done to make the strategies easier to use?
	What (if anything) could be done to make the program more convenient to use?

Data Analysis

Sociodemographic data and quantitative ratings from the BPI and the TAP questionnaire were analyzed descriptively. Frequency tables, medians, and ranges were used to summarize data for each item and to calculate the total score on the TAP measure. Data were not normally distributed (Shapiro-Wilk test<.05). Nonparametric Mann-Whitney U tests and chi-square tests were used to compare groups. Interviews were numerically recorded and transcribed by a qualified audio typist prior to content analysis following the approach by Miles and Huberman [34]. The QDA miner (Provalis Research) software was used to facilitate data management and organization of codes. The PI and a research assistant (RA) completed a 2-day training session on the use of this software. A preliminary generation of codes was based on the following attributes of acceptability highlighted by Sidani et al [19,33]: appropriateness, effectiveness, suitability, and willingness to adhere (see Table 1). Descriptive codes were created by attributing a code to each unit of analysis (words, phrases, or paragraphs). To ensure rigor and enhance credibility, separate coding (double coding) was conducted by the PI and the RA for the first 5 women and the first 5 men. Results were compared, and differences were discussed until a consensus was reached. However, no major differences were found. Additional codes (subcategories) were created when necessary. When a new code was generated, it was discussed as well. The PI kept a diary and noted questions or ideas and the discussions that occurred throughout the entire analysis process. Merging of similar descriptive codes created thematic categories representing a set of conceptual components. The content of each category and subcategory was compared between men and women. Frequency counts were also done to validate the emergence of a difference between the 2 subgroups.

Results

Sample Description

Eligible participants were recruited between November 2012 and May 2013. Characteristics of the sample are presented in Table 3. Participants ranged in age from 36 to 74 years (women, 36-74 years; men, 50-72 years). The majority of participants lived with a significant other except for 3 women and 2 men that were either divorced/separated or widowed but had a family member or a friend living close by. Of the 10 men, 7 had university degrees and 6 were still working. The chi-square test was significant for employment status (χ^2_1=4.9, *P*=.03). Most participants (16/20, 80%) had undergone a coronary artery bypass graft (CABG). It is noteworthy that almost 50% (9/20, 45%) of participants suffered from chronic pain—noncancer and noncardiac pain—with a median duration of 13 years for women (ranged from 72-360 months) and 3 years for men (ranged from 12-240 months). However, the Mann-Whitney U test was not significant for chronic pain duration (U=3.5, *P*=.11), meaning that there was no difference in chronic pain duration between women and men.

Table 3. Sociodemographic characteristics of participants.

Variables		Women (n=10)	Men (n=10)	Total (n=20)
Age in years, median (range)		67.0 (36-74)	65.5 (50-72)	66.5 (36-74)
Marital status, n (%)				
	Single	0 (0)	0 (0)	0 (0)
	Married or free union	7 (70)	8 (80)	15 (75)
	Separated/divorced/widowed	3 (30)	2 (20)	5 (25)
Living arrangements, n (%)				
	Lives with spouse (with or without children)	2 (20)	2 (20)	4 (20)
	Lives with family member or friend	5 (50)	6 (60)	11 (55)
	Lives alone	3 (30)	2 (20)	5 (25)
Education level, n (%)				
	Elementary school	1 (10)	0 (0)	1 (5)
	Middle school	5 (50)	2 (20)	7 (35)
	High school	2 (20)	1 (10)	3 (15)
	University	2 (20)	7 (70)	9 (45)
Employment status, n (%)				
	Full time/part time	2 (20)	6 (60)	8 (40)
	Retired	8 (80)	4 (40)	12 (60)
Presence of chronic pain, n (%)		4 (40)	5 (50)	9 (45)
Duration of chronic pain in months, median (range)		156.0 (72-360)	36.0 (12-240)	72.0 (12-360)
Type of surgery, n (%)				
	CABG	8 (80)	8 (80)	16 (80)
	Valve replacement (VR)	1 (10)	2 (20)	3 (15)
	CABG + VR	1 (10)	0 (0)	1 (5)

Postoperative Pain Intensity and Interference

All patients had experienced postoperative pain following discharge in the previous month. Most participants (18/20, 90%) reported that the pain had decreased since discharge, except for 2 men who reported that it had resolved. In terms of pain frequency, 6 participants—3 men and 3 women—reported that pain was continuously present, 12 participants—6 men and 6 women—reported that it was occasionally present, and 2 men stated it was absent. Women reported a higher, but still mild, level of average pain (Table 4). Using the Mann-Whitney U test, a statistically significant difference was found solely for worst pain (U=19, *P*=.03). Women reported a higher intensity of worst pain, which was moderate compared to mild for men. However, when looking at pain interference with activities, women experienced less pain interference with breathing and coughing than men (U=20, *P*=.04).

Table 4. Median (minimum-maximum) of pain intensity and interference with activities in the last 7 days, according to the BPI.

Item	Women (n=10)	Men (n=10)	Total (n=20)	*P* value (Mann-Whitney U test)
Pain now	3.0 (0-5)	0.0 (0-6)	2.0 (0-6)	.31
Average pain	4.0 (2-5)	2.5 (0-4)	3.0 (0-5)	.08
Worst pain	6.0 (3-8)	3.5 (2-8)	5.0 (2-8)	.03
General activity	4.0 (0-9)	2.0 (0-8)	2.0 (0-9)	1.0
Mood	0.0 (0-5)	3.5 (0-7)	2.0 (0-7)	.40
Walking	0.0 (0-5)	0.0 (0-6)	0.0 (0-6)	.40
Work	2.0 (0-9)	0.0 (0-8)	0.0 (0-9)	.63
Relationships	0.0 (0-5)	0.0 (0-4)	0.0 (0-5)	.97
Sleep	4.0 (0-8)	1.0 (0-7)	1.0 (0-8)	.84
Enjoyment	0.0 (0-5)	0.0 (0-5)	0.0 (0-5)	.90
Appetite	0.0 (0-7)	0.0 (0-8)	0.0 (0-8)	.72
Concentration	0.0 (0-6)	0.0 (0-5)	0.0 (0-6)	1.0
Breathing/coughing	0.0 (0-6)	3.0 (0-8)	2.0 (0-8)	.04

Acceptability of the SOULAGE-TAVIE Intervention

Overview

Participants' total scores on the TAP measure indicated that SOULAGE-TAVIE is very acceptable. No difference was found in their overall appreciation. The results for each of the four attributes of the TAP measure is presented for men and women in Table 5, followed by a summary of the qualitative data that was generated from the interviews. The differences between men and women are described if necessary.

Table 5. Median (minimum-maximum) of ratings for intervention attributes according to the TAP measure.

Intervention attributes	Women (n=10)	Men (n=10)	Total (n=20)	*P* value (Mann-Whitney U test)
Effectiveness				
How effective do you think the program would have been in helping you manage pain after cardiac surgery?	4.0 (3-4)	3.0 (2-4)	3.0 (2-4)	.06
How effective do you think the program would have been in helping you decrease the impact of pain on your recovery?	3.0 (2-4)	3.0 (2-4)	3.0 (2-4)	.50
Appropriateness				
How acceptable/logical does the program seem to you?	4.0 (3-4)	3.0 (2-4)	4.0 (2-4)	.008
How appropriate does the program seem to be to help with pain management after surgery?	4.0 (3-4)	3.0 (2-4)	3.0 (2-4)	.28
Suitability				
How easy does it seem to use the program?	4.0 (3-4)	3.5 (3-4)	4.0 (3-4)	.84
How easy do you think it would have been for you to apply all strategies?	4.0 (3-4)	4.0 (3-4)	4.0 (3-4)	.90
Willingness to adhere				
How willing would you have been to use the program?	4.0 (2-4)	4.0 (2-4)	4.0 (2-4)	.90
Total	3.6 (2.9-4.0)	3.3 (2.6-4.0)	3.4 (2.6-4.0)	.28

Experience of Raising Awareness

Overview

The first category that emerged after participants commented on their ratings of SOULAGE-TAVIE's effectiveness and appropriateness was experience of awareness raising. Participants' responses on the TAP measure in terms of effectiveness indicated that the intervention would have been very effective in helping them to manage their pain after surgery and in decreasing the impact of pain on their recovery. Women rated effectiveness slightly higher than men, although the difference was not statistically significant (U=22.5, *P*=.065). During the interviews, the benefits highlighted by participants were often related to satisfying their emotional needs, as

reflected by the following interview excerpts: "What the nurse says is reassuring," "You know what to expect," and "It tones it down." Informational benefits of SOULAGE-TAVIE were also highlighted by the following excerpts: "Before surgery you don't necessarily ask the good questions," "I would have known what to do," and "You hear so many things from people before surgery, it gives you a clear answer." Regarding appropriateness, participants rated the intervention as very logical and appropriate for pain management. A statistically significant difference was found for the question "How acceptable/logical does the program seem to you?" (U=13.5, *P*=.008), with women scoring higher than men. In the qualitative comments, appropriateness was related to the timing of the intervention and relevancy of the advice, as reflected by the comment "It would have been great to have this (intervention) before surgery." Many participants stated, "All advice is important."

Awareness raising emerged as both women and men outlined benefits that extended beyond information and reassurance. An awareness-raising experience was mentioned more often than emotional and informational benefits and was related to two subcategories: awareness raising toward pain beliefs and awareness raising toward pain management behaviors.

Experience of Awareness Raising Toward Pain Beliefs

When explaining how the intervention increased their awareness toward pain and its relief, women stated that they had been afraid to take pain medication, but now understood how pain medication could be helpful to their recovery, as reflected by the following excerpt: "I didn't think I was so scared of pain medication." However, men became aware that their beliefs about pain were dated and that it was important to relieve pain, as reflected by the following excerpts: "It's normal to experience pain after surgery…you put your prejudices aside," and "Often we have preconceptions…you end up punishing yourself." Another man evoked his beliefs: "I wasn't enough aware of the importance of relieving pain…I was educated the old way."

Experience of Awareness Raising Toward Pain Management Behaviors

While referring to their experience of awareness toward pain beliefs, participants tended to reflect on their pain management behaviors. One man mentioned that the approach was innovative: "It makes you think…I didn't have the right behavior toward pain—I would have acted differently." Another man commented on his pain management behavior: "I tried to avoid moving too much." A woman expressed her awareness-raising experience toward pain relief as follows: "Just to be aware that pain can be treated…it gives you the opportunity to be proactive. Sometimes I was waiting a long time before doing something." Finally, all participants referred to the most important advice they had retained, which was almost always "to not put up with pain" or "to avoid peaks." A good number of participants summarized the advice as "being preventive toward pain."

Experience of Convenient Support

Overview

The second category that emerged after having participants comment on their rating of SOULAGE-TAVIE's suitability was experience of convenient support. Participants reported that the intervention was very suitable. The program was rated as very easy to use, and the strategies were rated as very easy to apply. Ratings on the TAP measure were almost identical for women and men. When commenting, more men than women reported that they were comfortable with the Web-based delivery mode. Some women were a bit reticent toward the use of the computer at the beginning of the interview but evolved during the demonstration and discussion, as reflected by the following interview excerpts: "It would have been a bit difficult at the beginning but I would have managed…I could have asked my grandson," "I don't use computers but I would have tried," and "Now that you showed it to me it's not complicated." The majority of participants commented on suitability in terms of adequate length, timing, and, especially, delivery mode. The use of a Web-based application as a delivery mode was perceived as a logical fit with today's living, as reflected by the following excerpts: "Everyone goes on the Web to look for information," "It's a new era," and "Nowadays, computers are essential." Two subcategories emerged while describing their experience of support: flexibility and interaction.

Experience of Flexible Support

Flexibility in terms of personal readiness was the most reported advantage of using a Web application, as reflected by the following excerpt: "You can use it at your convenience…with a clear head." Flexibility was also illustrated in terms of improved access to information in the context of acute care by the following excerpts: "You can go back anytime," and "If the nurse could not take the time to explain, I'd still have this information."

Experience of Interactive Support

The virtual nurse was another aspect raised with regard to the delivery mode. A new category was then added as participants identified the benefits of interacting with the virtual nurse. Participants described two aspects related to the interaction. First, they perceived they were more attentive and they had a better retention of the information as reflected by these excerpts: "It's easier to understand when you listen to the nurse," "You are more focused…sometimes, when you read you don't do it right, you want to finish quickly," and "She catches your attention." The other aspect is more related to the relationship with the nurse: "It's more friendly," "I find it more personalized…you feel like you're talking to someone," "It's not like being in front of a real nurse but it's more human…it breaks the ice," "I trust her," and "It adds some authenticity to the advice."

Experience of Guidance

The third category that emerged after having participants comment on their ratings of their willingness to adhere to SOULAGE-TAVIE was experience of guidance. Participants' scores on the TAP measure for men and women indicated that they were very willing to use the Web application. Similarly, during the interviews they insisted that they would have liked to use the Web application before their surgery: "It would have been useful," "It would have been good for me," and "It's a good program." Men and women were very satisfied with their experience with the intervention. It is noteworthy that participants tended to refer to the program as a "good guide"

allowing for self-determination and control: "Advice remains advice, it is not an obligation…you cannot take people by the hand." Men expressed their willingness to adhere to the intervention by pointing out the suitability of the intervention: "It's a useful tool…very practical." Women tended to focus on the importance and relevance (appropriateness) of the advice for other patients facing pain after cardiac surgery: "Necessary advice that everyone should get," and "It's important that everyone knows." Table 6 summarizes the content analysis for both women and men.

Table 6. Content analysis summary for women and men.

Attributes of the TAP measure	Category	Subcategory	Representative quotes from groups, verbatim
Effectiveness: In helping manage pain In helping decrease pain impact	Awareness raising	Toward pain beliefs	Women, on medication reluctance: "I didn't think I was so scared of pain medication." Men, on pain normalization: "Often we have preconceptions…you end up punishing yourself."
Appropriateness: Logical/acceptable Appropriate to help with pain management	Awareness raising	Toward pain management behavior	Both: "It makes you think…I didn't have the right behavior toward pain, I would have acted differently." Both: "Just to be aware that pain can be treated…it gives you the opportunity to be proactive—sometimes I was waiting a long time before doing something."
Suitability: Easy to use Easy to apply	Convenient support	Flexible	Both, on readiness: "You can use it at your convenience…with a clear head." Both, on access: "You can go back anytime."
		Interactive	Both, on attention: "You are more focused." Both, on interaction: "It's more friendly."
Willingness to adhere	Guidance	Self-determination	Both: "It's a good guide." Both: "You can't take people by the hand." Women, on awareness raising: "It's important that everyone knows." Men, on convenient support: "It's a useful tool…very practical."

Discussion

Principal Findings

The aim of this study was to provide a thorough description of the acceptability of a Web-based intervention for the self-management of pain after cardiac surgery and to delineate potential differences between the perceptions of women and men about the acceptability of the intervention. Based on the TAP scores, the intervention was perceived as very acceptable by both groups. Although the sample was quite small, it was observed that women rated the intervention higher in terms of appropriateness than men. Women's higher postoperative pain intensity scores and longer duration of chronic pain may have resulted in greater support needs and, consequently, they experienced more satisfaction with the intervention. This difference might also be related to psychosocial characteristics. It was previously observed that women experienced emotional distress while waiting for surgery and after discharge and that they sought to "preserve the self" and accept their modified health and functional status [35-37]. Moreover, in the current study more women were retired than men. Hence, the mixed-method approach allowed for a better understanding of the TAP measure scores—total and specific attributes—since they were high and quite homogenous across men and women.

Overall, the participants indicated that in addition to the emotional and informational benefits, the SOULAGE-TAVIE intervention would have generated an experience of increased awareness of their own pain beliefs and pain management behaviors as well. Furthermore, men and women underlined that they would have acted differently in the face of pain. This finding is very interesting because it validates the appropriateness of the underlying therapeutic strategy and the content of messages, and demonstrates the acceptability of the method for patients experiencing postoperative pain after cardiac surgery. Indeed, the intervention development was based on the Elaboration Likelihood Model (ELM), which focuses on imparting information that stimulates reflection and a change in attitude [38,39]. If motivated, individuals are active information processors as they can carefully consider messages, and relate them to other information and to their own experiences [39-41]. The success of this strategy relies on the selection of appropriate tailoring variables that will enhance message relevance (ie, pain belief and attitudes) but also influence the targeted behavior (eg, pain management). Hence, the use of theory and the combination of behavioral change techniques were associated with an increase in impact by Web-based interventions [12]. The only difference between women and men was the nature of this awareness raising. Women reported awareness about their reluctance to use medication and tendency to wait too long to use pain medication. This is consistent with previous work that found women wanted to take as little medication as possible and did not follow the recommendations despite reporting high levels of pain [22]. Men reported awareness about their preconceptions and normalization of pain, which led to the same behavior of procrastinating before relieving their pain. In fact, the computer tailoring method, which screens for pain beliefs and attitudes before message delivery, addresses this gender difference regarding awareness.

The second main result is that participants would have experienced a convenient support. Men and women judged the Web-based intervention as suitable because of its flexibility. Indeed, Web-based, tailored interventions seem to offer both more control to patients in terms of content and timing [14,24], and more outreach when access to care is limited [42,43]. According to the participants, the Web-based delivery seemed to include another advantage over a more traditional printed format: increased attention and retention. They found it was easier to integrate the information and stay focused. Indeed, Web-based tailored interventions provide greater interactivity and may result in more engagement [14]. In fact, the virtual nurse personified interactivity in SOULAGE-TAVIE and personal contact tends to support behavior change in Web-based interventions [12]. Not only does the use of information technologies allow messages to be more attractive [24], but it also seems to enhance cognitive processing through customization [44,45]. An experience of guidance accompanied by self-determination rounded out their overall appreciation of the intervention. This result underlines the acceptability of the general approach of SOULAGE-TAVIE in terms of user control—self-guided/automated as opposed to expert led/directed—for this population [24]. This type of approach is usually privileged with brief interventions [24]. Finally, it is noteworthy that men expressed their willingness to use or adhere to the intervention due to format suitability and practicality, whereas women cited the appropriateness of advice. Until now, gender has not been shown to make a difference in terms of interest in Web-based, delivered interventions [43] or effect size [11,14]. Nevertheless, education might have played a role in this difference of perception considering that fewer women had a university level of education. Indeed, it was previously demonstrated that lower-educated individuals showed higher attention and processing of information which led to greater intention to use the website [46].

Limitations and Future Directions

As with other studies, this study had some limitations. The participants were recruited approximately one month after their cardiac surgery. They were not in a preoperative frame of mind, which may have influenced their perceptions with regard to effectiveness and appropriateness. However, the recent experience of surgery and postoperative pain may have contributed to the perceived relevance of the intervention. Also, more women were retired and could have suffered from social isolation, which could contribute to the appreciation of the intervention and the study participation.

However, the participants' experience with the intervention during the recovery phase revealed the relevance of the intervention before surgery and after discharge as well. Previous studies on pain after cardiac surgery following discharge highlighted high pain levels and the contribution of pain beliefs and attitudes to pain levels, especially in women [21,22]. Authors highlighted the relevance of an intervention initiated before surgery and extended after discharge [47]. Moreover, the delivery of information at discharge seems to face the same issues as the delivery of information before surgery [47]: patients have little time to integrate content. Thus a Web-based intervention could be a good option to face the challenges experienced both preoperatively and during the recovery period. Considering that SOULAGE-TAVIE has demonstrated potential effects on the pain experience in the first week after surgery, it would be interesting to assess its impact after discharge and explore the possibility of preventing chronic pain after cardiac surgery or at least disability. Moreover, SOULAGE-TAVIE targets psychosocial characteristics that are included in models looking at the transition from acute to chronic postoperative pain [5,48].

Conclusions

When designing a new intervention, the mixed-method approach and data triangulation are very useful as they capture patients' perceptions and the mechanisms underlying the effectiveness of intervention. This study described the acceptability of computer tailoring and persuasive communication to modulate pain beliefs and attitudes in an acute care context. Participants perceived the intervention to be very acceptable in terms of content and format, although women and men differed in their reasons for its acceptability. Women were willing to adhere to the intervention based on the importance and relevance of the advice provided whereas men were more focused on the delivery mode and their appreciation of its flexibility. The participants' experience of SOULAGE-TAVIE after discharge revealed its relevance across the continuum of care. This approach seems to be an interesting avenue to influence the transition from acute to chronic postoperative pain.

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Conflicts of Interest

None declared.

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Abbreviations

BPI: Brief Pain Inventory
CABG: coronary artery bypass graft
CPOP: chronic postoperative pain

ELM: Elaboration Likelihood Model
PI: principal investigator
RA: research assistant
RCT: randomized controlled trial
SOULAGE-TAVIE: SOUtien à L'AutoGEstion-Traitement-Assistance Virtuelle Infirmière-Enseignement
TAP: treatment acceptability and preference
VR: valve replacement

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Original Paper

Tying eHealth Tools to Patient Needs: Exploring the Use of eHealth for Community-Dwelling Patients With Complex Chronic Disease and Disability

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Abstract

Background: Health policy makers have recently shifted attention towards examining high users of health care, in particular patients with complex chronic disease and disability (CCDD) characterized as having multimorbidities and care needs that require ongoing use of services. The adoption of eHealth technologies may be a key strategy in supporting and providing care for these patients; however, these technologies need to address the specific needs of patients with CCDD. This paper describes the first phase of a multiphased patient-centered research project aimed at developing eHealth technology for patients with CCDD.

Objective: As part of the development of new eHealth technologies to support patients with CCDD in primary care settings, we sought to determine the perceived needs of these patients with respect to (1) the kinds of health and health service issues that are important to them, (2) the information that should be collected and how it could be collected in order to help meet their needs, and (3) their views on the challenges/barriers to using eHealth mobile apps to collect the information.

Methods: Focus groups were conducted with community-dwelling patients with CCDD and caregivers. An interpretive description research design was used to identify the perceived needs of participants and the information sharing and eHealth technologies that could support those needs. Analysis was conducted concurrently with data collection. Coding of transcripts from four focus groups was conducted by 3 authors. QSR NVivo 10 software was used to manage coding.

Results: There were 14 total participants in the focus groups. The average age of participants was 64.4 years; 9 participants were female, and 11 were born in Canada. Participants identified a need for open two-way communication and dialogue between themselves and their providers, and better information sharing between providers in order to support continuity and coordination of care. Access issues were mainly around wait times for appointments, challenges with transportation, and costs. A visual depiction of these perceived needs and their relation to each other is included as part of the discussion, which will be used to guide development of our eHealth technologies. Participants recognized the potential for eHealth technologies to support and improve their care but also expressed common concerns regarding their adoption. Specifically, they mentioned privacy and data security, accessibility, the loss of necessary visits, increased social isolation, provider burden, downloading responsibility onto patients for care management, entry errors, training requirements, and potentially confusing interfaces.

Conclusions: From the perspective of our participants, there is a significant potential for eHealth tools to support patients with CCDD in community and primary care settings, but we need to be wary of the potential downfalls of adopting eHealth technologies and pay special attention to patient-identified needs and concerns. eHealth tools that support ongoing patient-provider interaction, patient self-management (such as telemonitoring), and provider-provider interactions (through electronic health record integration) could be of most benefit to patients similar to those in our study.

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KEYWORDS

eHealth; primary health care; patient-centered care; chronic disease; multimorbidity

Introduction

Health systems globally are shifting attention towards examining high users of the health system. In Ontario, Canada, only 1% of the province's population accounts for 34% of costs, while 10% accounts for 79% of total system-wide costs [1]. Similar trends are also found in British Columbia, Canada [2], and in the United States [3]. The small group of high users includes a number of subpopulations; among them are patients with complex chronic disease and disability (CCDD). Patients with CCDD can be characterized as having multimorbidity (having two or more chronic illnesses) [4] and symptoms that have an impact on their daily living [5], which results in their using more care [1,6,7], experiencing poor care coordination [8], and having a higher risk of poor health outcomes than those with single illnesses only [7,9]. Biology and disease profile, however, capture only the chronic disease and disability aspect of CCDD. The complexity aspect requires attention to broader social, environmental, and contextual issues that have an impact on the health care needs of these patients, leading some to call for patient-centered approaches to care delivery [4].

Patient-centered care requires a "focus on the patient's experience of illness and health care and on the systems that work to meet individual patients' needs" (p. 48 [10]). A patient-centered approach to care requires focus at multiple levels. At the patient-provider level, patient-centered care involves communication, respect for patients, shared responsibility between patients and providers, access to information and education for patients and families, and support for the whole patient (ie, from a bio-psychosocial perspective). At the system level, patient-centered approaches require organizations and systems that place the patient at the center of care with particular attention to coordination, integration, and continuity of care [10-14]. eHealth technologies may be a key strategy to supporting patient-centered care through their ability to support improved access, continuity, communication, shared decision-making, and patient self-management [15-19].

While there have been many advances in adopting eHealth technologies to support chronic disease patients in hospital settings [20] and primary care settings [21], many of these tools are disease specific and may not be able to address the needs of patients with CCDD. We sought to address this gap by developing a suite of eHealth mobile apps and tools for use in team-based primary care settings to support patients with CCDD living in the community. In our broader project, we used a design evaluation approach that involves refining designs based on prior research and ongoing evaluation that involves end-users throughout the process [22]. In this paper, we report on the first stage in our development process in which we use an interpretive descriptive qualitative methodology to identify the perceived needs of community-dwelling patients with CCDD with respect to (1) the kinds of health and service issues that are important to them, (2) the information that should be collected and how it could be collected in order to help meet their needs, and (3) their views on the challenges/barriers to using eHealth mobile apps to collect the information.

Methods

Research Design

An interpretive description approach [23,24] was used to guide our study design and analysis method. Interpretive description, which comes from qualitative nursing research, aims to describe and interpret a "shared health or illness phenomenon from the perspective of those who live it" (p. 171 [23]). Given our intention to better understand the perceived needs of community-dwelling patients with CCDD, and the information sharing and eHealth technologies that could support those needs, an interpretive descriptive design was determined to be an appropriate approach.

Context

Focus group participants were recruited from a Family Health Team (FHT)—an interprofessional primary care delivery model [25] in Ontario, Canada. The practice serves over 5000 people from the Riverdale community of Toronto as well as the Greater Toronto Area. The FHT is composed of 22 staff members: 6 primary care providers, 1 social worker, 2 registered nurses, 2 medical assistants, 3 diabetes educators, and 8 administrative staff.

Sampling and Recruitment

Focus groups were conducted with community-dwelling patients with CCDD to learn what kinds of health and service issues were important to participants, what information should be collected, and how it could be collected in order to meet their needs. Purposive criterion sampling [26,27] was used to identify community-dwelling patients with CCDD to participate in this phase of our study. Purposive sampling is an appropriate approach for interpretive description studies like ours [24].To be included, focus group participants had to (1) have been identified as a patient with CCDD (defined as individuals with one or more health conditions that are difficult to manage), (2) be a patient at the FHT, (3) have the ability to give informed consent, and (4) understand and speak English. Approximately one third of the 5000 FHT patients fell into our definition of CCDD. Eligible participants were identified with the help of FHT staff. Recruitment posters with eligibility criteria, researcher contact information, and a brief description of the study were posted in the designated waiting area of the FHT as well. Participants provided consent to FHT staff to share their contact information with the research team and/or participants contacted research team staff directly to be included in the study. In a couple of cases, the patient was accompanied by their caregiver who expressed interest in participating and/or was required to attend to provide assistance to the patient. We did

not originally intend to include caregivers, but those who expressed interest in participating were invited to attend.

Procedure

Focus groups took place between November and December 2013. Between 6 and 9 participants were assigned to each focus group based on availability. After providing consent, all participants filled out a "participant information sheet", which was used to collect data on age, gender, country of origin, and chronic illness profile. The catchment area of the FHT serves a diverse population of high and low income residents, and so we anticipated capturing a diverse group with regard to socioeconomic status. We did not feel the need to formally gather socioeconomic status data such as occupation or household income. Focus groups were semistructured around the questions listed in Textbox 1.

In addition to these questions, participants also had the opportunity to try out an example of a mobile monitoring system using a tablet, after which participants were asked: "What was it like answering questions using a tablet? What did you think about the content and wording used in the questionnaire that was downloaded onto the tablet?"

Focus groups lasted between 90 and 120 minutes and were audio recorded and transcribed by an external source. Transcripts were checked by the lead author (CSG) for accuracy. In addition to answering questions, participants were also presented with an example of an eHealth mobile app. Participants were invited to discuss whether a tool similar to the example provided might meet their needs and what types of challenges/barriers they may experience in using this type of technology.

Focus groups were conducted until new data resulted in only minor variations on identified themes in the codebook (ie, thematic saturation) [28,29]. Analyses of the first three focus groups generated a set of themes that were unchanged by the fourth focus group. As such, we were confident that nothing new could be learned from additional focus groups. An inductive analytic process that seeks thematic saturation is appropriate for interpretive descriptive research designs.

Textbox 1. Focus group questions.

1. We are interested in understanding your experience in the health care system.
   - Can you share with us the things that are important to you as a receiver of health care services?
   - What can be done to improve things?
2. It is important for the health care system to gather information from you to better understand you and improve your care.
   - What type of information should be collected from you?
3. How can health providers (or you as a person who uses health care) use technology to collect this information?

Data Analysis

Inductive analysis was conducted concurrently with data collection through the identification, discussion, and notation of prominent themes between the two researchers conducting each focus group, generating a preliminary codebook that was applied to one focus group transcript by three researchers (CSG, DM, and CC). The prominent themes were discussed by the research team, and the codebook was revised.

Using the revised codebook, 2 researchers (CSG and DM) independently coded all transcripts using QSR NVivo 10 software. After each transcript was coded, the 2 researchers compared coding and reached consensus on all codes, modifying the codebook and codes applied to the transcript to reflect the consensus that was reached. For example, there was a discrepancy between how the concept "patient as expert" was coded by the 2 researchers, mainly revolving around whether patients viewed themselves as an expert in their care or perceived that the provider viewed them as an expert in their care. After reviewing a second and third transcript, the researchers came to a consensus that the concept should include both ideas (self-perceived and perception of the provider viewing the patient as an expert). The codebook was then modified and transcripts re-coded to reflect the new definition of the code.

This process was followed for each of the four transcripts. By the third and fourth transcripts, there were few discrepancies between the 2 researchers, demonstrating reliability of the thematic coding. The 2 researchers identified emerging subthemes through the coding process that are included in the findings. For example, the code "patient identified area of importance - communication" code applied to communication between patient and provider, between providers, and could include multiple forms of communication (ie, in person, telephone, electronic). These subthemes were identified by the 2 coders through the coding process to tease out the broad concept of communication. The coded data were next analyzed to identify relationships between codes. A table was created to demonstrate coding overlap, which was discussed and agreed on by the entire research team. This table informed the creation of an illustrated framework that demonstrates the connections between themes and subthemes. The framework is presented in the results section of this paper. The use of visual tools like our framework are recommended as part of the interpretive description approach [24] and help us to clarify how our concepts are related to each other.

In order to test the trustworthiness of the data, all focus group participants were given the opportunity to review the findings and provide feedback to the research team. Findings were presented in terms of concepts and themes representing the entire sample. This is an appropriate approach to participant validation for an interpretive descriptive study [23]; 6 of the 14

participants were amenable. The findings summary was mailed along with a feedback form and self-addressed and stamped envelope for the participants to fill out and return. Three responses were returned and confirmed that findings reflected their experiences and those discussed in their respective focus groups. Debriefing activities like this serve to support the credibility and trustworthiness of the data analysis [23,30]. It should be noted that one respondent identified additional subconcepts within the codes that were mentioned in their focus group, but that the participants felt were not evident in the summary. The subconcepts were reflected in the more detailed analysis used by the research team and as such were still captured in the analysis.

Results

Participants

The focus groups were conducted with patients with CCDD (n=10), caregivers (n=2), and those who were both caregivers and patients with CCDD (n=2). Patients included in the focus groups reported having multiple chronic illnesses including diabetes, chronic pain, osteoarthritis, osteoporosis, anemia, cardiac conditions, glaucoma, and mental illness. The average age of participants was 64.4 years; 9 participants were female, and 11 were born in Canada. While education level, socioeconomic status, and technological aptitude were not formally captured, these data were captured through researcher observation as well as through the information shared by participants during the focus groups. All participants were able to read and understand the consent form, which suggests at least a moderate literacy in English. Through the focus group conversations it was made clear that nearly half (n=6) held professional jobs that would require at least some post-secondary education. Most participants expressed that they were comfortable with computers and smartphones when they were presented with a device. Four participants made it clear that they were not as comfortable with these forms of technology, but only one participant did not attempt to engage with the sample device provided at the session.

Each focus group had between 2 and 5 members. Although our original aim was to have between 6 and 9 participants as suggested in the literature [31], but there were a number of last minute dropouts mainly due to illness. The timing of the dropouts did not allow for rescheduling within project timelines, and we did not wish to further burden patients by asking them to return. Hence, focus groups were conducted as per the original schedule. While the concern with low numbers in focus groups is a lack of adequate discussion [31], this was not a problem in any of the focus groups. Given that we reached thematic saturation (described above), the research team determined that additional groups with more participants were not required. Quotes from participants are identified by the focus group in which they participated.

Important Issues for Patients With Complex Chronic Disease and Disability Receiving Health Care Services

Patient-Provider Interactions

Participants assigned high importance to their interactions with their primary care providers at the FHT, specialists, pharmacists, nurses, and health care administrators. Of importance to participants was the need for open, ongoing, two-way communication between themselves and their providers, particularly around test results:

> *I also need to know the results of tests when they happen. I need to know them…Like I need to look back and say this is what your test did, this is what it revealed, and this is what it means for the futureAnd if something is prescribed for me, why am I getting it or why is [my spouse] getting it, and what's it supposed to do? And if it doesn't do it, what do we do?* [FG 1]

Some participants noted that this timely feedback could help them to manage anxiety they were experiencing regarding their health:

> *So I go down and get the ultrasound [to check a lump]. And it was a good 5, 6 days before we get the information. It turned out to be nothing. But in those 5 days, I'm sitting there thinking, you know, have I got it [cancer]…It really works on your mind, you know.* [FG 2]

Participants did not just want to share information back and forth, but they wanted that exchange to be of high quality. Participants wanted an open "dialogue" with their providers in a space where they felt "heard" by a provider who was "taking time" to respectfully listen:

> *I don't care if it's on the phone, in-person, just make the time. Don't rush us out the door like we're a bloody number. We're not on the slab, you know. We're not a piece of meat. Listen to us, deal with us. Don't push us out the frigging door because you're not helping us like that.* [FG 3]

However, participants were weary of having to repeat themselves to different providers and of feeling as though they needed to "start from scratch" with every new provider they saw. Participants saw this as an issue that could be addressed through better information sharing between providers. Some participants suggested that improved patient information sharing between providers could be a proxy for ongoing relationships with a single provider that knew the patient's history.

Provider-Provider Interactions

As might be expected, patients with CCDD tend to have multiple providers. Coordinating care between these providers was identified by participants in all focus groups as an ongoing problem in their care with regard to ensuring appropriate referrals, medication management, visits to the hospital, and overall coordination of care. One story provided by a participant describes the communication breakdown between a hospital and primary care provider:

> *That hospital did not notify [my primary care doctor]…I got out of the hospital and [my primary care doctor] said to me, "What happened?" I said, "Well, I don't know what happened but I had to have bowel surgery."…They did not give her any info on me. And she's my family doctor.* [FG 3]

One participant noted a key issue with the lack of communication is that no one was looking at them holistically, stating:

> *…what happens is you have…somebody who looks at your hand, somebody who looks at your head. And nobody connects the whole thing together.* [FG 4]

An important issue raised by participants was identifying a single provider who had responsibility for the management of their information. Participants saw their primary care provider as being the "gatekeeper" of their information and as being responsible for having comprehensive information gathered from all providers:

> *If they're specialists, they have limited knowledge and they don't need to know about everything else about you. And I think it's important that the family doctor communicate with the [specialist]. That the family doctor should be the gatekeeper of your charts and your data.* [FG 4]

> *And it's true for all of us, if we don't have a primary care physician who's coordinating and navigating all of that, and helping us to understand what it is, then we're off…through the system.* [FG 1]

Participants also wanted to know when their providers were communicating with each other, demonstrating a desire to be a part of the care process:

> *It's done as though we're not really a part of it. So until the second doctor gets back to us with an appointment, we have no knowledge whether the first contacted them or not. So it's like if it's supposed to take 4 months, we have to wait 4 months. And if it goes to 5 months, 6 months, then we might find out that they never did it. We're left out of the equation.* [FG 3]

Access

A third key issue for participants was access to needed health care services, specialists, and treatments (mainly medications). Analysis revealed that access issues were often related to the patient-provider interaction issues identified above. Participants reported waiting to hear back from providers, to see providers (ie, in waiting-rooms), and for hospital beds. One caregiver thought that keeping patients waiting for a long time showed disrespect towards patients:

> *I want my time valued. I don't want to sit in an office with my partner who is pretty hard of hearing, not deaf, and has complex health problems, watching him get more and more uncomfortable. And actually I'm very happy with the fact that here they're seen promptly and it's organized promptly. And that tells me that people respect me. That kind of respect is incredibly important.* [FG 1]

Participants reported that limited mobility made transportation to and from appointments challenging and that the costs associated with uninsured services impeded access:

> *But the transportation, my issue was that when you go to a specialist, you want to be able to go to someone who is on [public transit] because parking is an arm and a leg everywhere you go. And if they keep you waiting then you're paying for 3 hours of parking.* [FG 4]

> *I don't want them to say, 'Well, here's a list of practitioners that you should contact and get some…' Like excuse me, I can't afford $120 a visit. I'm retired now. So I would really like people to ask what kind of benefits do you have, and have a real list. You know, if I can afford it, if it's covered, great. But if not, what resources are there for me beyond this relationship that I can avail myself of. Because to tell you the truth, and [spouse], my partner, that gives us a sense of hope and possibility. And we all need a sense of hope and possibility.* [FG 1]

A few participants shared concerns regarding inappropriate access, noting that they did not want to use scarce resources unless absolutely necessary. The fear of inappropriate use could actually deter patients from using services that may be needed, as illustrated by a story shared by one participant:

> *I had pain in my chest and in my jaw after I did some exercise. And I knew that these were no-no's …So I had to call the health line because they were the only people I could talk to. And when I described what I was feeling to the woman on the phone, she said, 'You go to [Toronto hospital] immediately. And you go to the emergency.' And I went there and they hurried me into a room where they kept me hooked up to all kinds of things all day long. And at 5:00 in the afternoon, they gave me dinner and they sent me home. And I felt relieved but I was also embarrassed because I was not having a heart attack. And I used up some precious time and tests and materials and space and doctors. So maybe the next time I feel that, I won't call. And maybe I will have a heart attack!"* [FG 1]

Patient-Centered Approach

Participants identified the need for a patient-centered approach to their care. As suggested in patient-centered care literature, participants described wanting to be treated as whole persons, to feel as though they are seen as experts in their own care, and they identified the need for a strong, ongoing relationship between themselves and their health care providers built on trust:

> *I had someone before who looked at me, pegging me immediately as someone like her mother…But you know, like no, I'm not your mother…You really need to start with respect. We all deserve respect. Don't*

have preconceived notions. Start with respect. Look at the whole person and really listen. [FG 1]

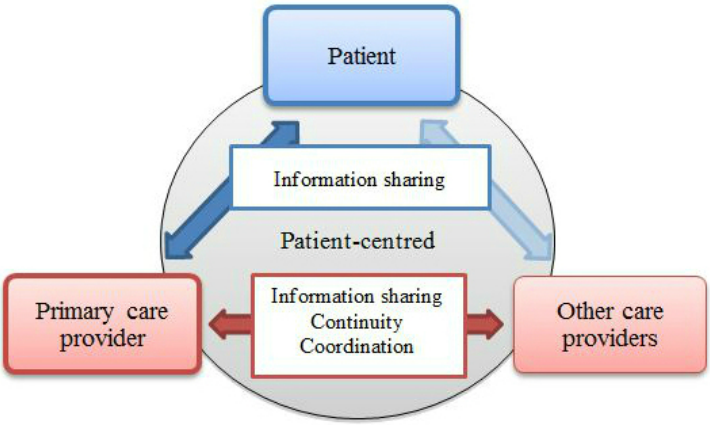
But also at a certain age, you do have a background of experience that says, you know, this is how your body is and you tend to swell up when you eat salty food. [laughs] So this is what happened…So I know these things. [FG 4]

It's my life and my health here. Make me a part of it. [FG 3]

Needs Framework for Community-Dwelling Patients With Complex Chronic Disease and Disability

The health care needs identified by the participants in our focus groups can be conceptualized using a relatively simple framework (see Figure 1). This framework helps to visualize the relationship of the different needs and to identify areas where we could focus our eHealth technology development to be of most use to our CCDD patients. We depict the information sharing, communication, and access using arrows. Placing the patient at the apex of the relationship is intended to support the notion of patient-centeredness, which was important to participants in our study. The arrow between patient and primary care provider is emphasized compared to the arrow between the patient and other care providers, reflecting participants' focus on their communication with their primary care providers and their feelings that their primary care providers are the central coordinators of their care. The arrows between providers are intended to reflect the need for bi-directional information sharing, continuity, and coordination identified as important to our participants.

Figure 1. Needs framework for community-dwelling patients with CCDD in our study.



Information Sharing to Improve Care Using eHealth Tools

Focus group participants were asked about what kinds of information sharing would best support their needs. With regard to information exchanged between themselves and their providers, participants identified wanting to share medical and medication history, information regarding their symptoms and other health outcomes, and experiences with care. In terms of provider-to-provider information sharing, the emphasis was on sharing of medical and medication history as a means of improving coordination and continuity.

Participants saw significant potential for eHealth technologies to support the needs they had identified, summarized in Table 1.

Table 1. Supporting our CCDD patient needs using eHealth technologies.

eHealth app	Purpose	Quotes
Patient-provider information sharing	Monitoring symptoms by provider and self- monitoring by patient	*…anything that can help replace another visit to the doctor or an easy way to be monitoring a person who's just come out of hospital at home, I think that it is so important.* [FG 1]
		You could set this up to keep track of just how much you're progressing or how much you're regressing. [FG 2]
	Patient accessing medical history	*But I'd want to know the results of the test.* [FG 3]
Provider-provider information sharing	Fast easy access to patient medical history	*I think the communication between each doctor would be a lot faster [using eHealth]. Like you'd have the patient file. They can each access it.* [FG 3]
	Coordination	*…if she was let go from the hospital, [the social worker] would have had all that information on the tips of their fingers—How is she going home and all, are we going to make something accessible, is the volunteer going to take her down? …Who is going to be at your home? Who is going to feed you? Do you want Meals on Wheels?* [FG 4]
	Continuity	*But besides that, it's in print right in front of the doctor. She can read it and know it's there, and she can recall it rather than, you know, talking on the phone with someone for 5 minutes and only taking in half of what the person said.* [FG 2]

eHealth Tradeoffs

While participants were excited about the potential for eHealth to support their ongoing needs, they also identified a number of concerns with using eHealth tools. Participants expressed concerns regarding privacy and data security, accessibility (visual or motor impairment issues affecting the use of smartphone and tablets), the loss of necessary visits, increased social isolation, a new burden for overstretched providers, downloading responsibility onto patients for care management, entry errors, training requirements, and potentially confusing interfaces. Many of the anticipated challenges were related to participant-identified advantages, suggesting that the selection and design of eHealth applications may warrant cost-benefit analysis and awareness of trade-offs.

For example, participants liked the idea of ongoing monitoring and avoiding unnecessary physician and hospital visits, but some expressed concerns that the use of eHealth technologies may displace necessary in-person visits or contribute to isolation:

> *Like for people who are like bedridden and can't get out, and you know, get their Meals on Wheels and stuff like that. If [eHealth monitoring is] the only contact that they're going to have, that's going to cut them off even more from society.* [FG 4]

Additionally while several participants called for wider sharing of patient information between providers, privacy and information security concerns were raised by others. Interestingly, a number of participants identified that the desire to have information shared easily, trumped their desire for privacy:

> *There are reams of x-rays and EKGs or ECGs. Stuff is sitting in doctors' offices. Therefore if I have to go to a new doctor for whatever the reason, I want them to have it all. Short and sweet. I don't care how. And I don't want it to be my decision.* [FG 1]

> *I would like any health care professional, a doctor, whether it's a specialist or a GP, be able to access that information.* [FG 4]

Discussion

Needs of Our Patients With Complex Chronic Disease and Disability

Our findings suggest that patients with CCDD at our FHT have a number of important care needs, among them being the need for improved communication and interactions between (1) themselves and their providers (both primary care and specialist providers), and (2) their different providers. Improved interactions between providers was also seen by participants in our study as a means to improve the coordination and continuity of their care. Our findings also highlight the need for these interactions recognizing the patient as a whole person and as an "expert" in their own care: concepts that are consistent with principles of person-centered approaches to care.

Findings from our study resonate with a previous study conducted with a similar patient population, but in an in-patient setting. Kuluski et al [32] conducted a qualitative study to help better understand the care needs and experiences of complex in-patients at a continuing complex care hospital in Toronto. The research team interviewed 116 patients who identified the need for improved communication with their providers and improved coordination of care (through supported transitions and more comprehensive patient assessment). A prominent theme in this study was the need for respectful interactions between providers and patients. Although the Kuluski et al study was conducted with in-patients, there are a number of similarities between the participants in this study and our own; the average age of participants was 63, mostly female, with multiple morbidities.

A number of ways that eHealth technologies could support the health care needs were identified by participants in our study. A key focus for our participants was the role eHealth technologies can play in supporting interactions between patients and providers and between different providers.

Developing eHealth Tools to Support Patient-Provider Interactions

Provider-patient interactions identified as important by participants involved patients sharing information back to providers regarding symptoms (monitoring) and patients' being able to access their health information. These communication pathways may be facilitated through the use of electronic health records, telemedicine or telehealth care, and technologies to support patient monitoring, sometimes referred to as telemonitoring [33-35]. Prominent eHealth tools that may be useful include:

- Electronic Medical Records (EMRs): Software used at a single organization to collect, manage, and store patient health information (replacing old paper files) [20].
- Electronic Health Records (EHRs): Electronic systems that allow for the sharing of health data across different providers and health organizations [36] (see also [37]).
- Electronic Patient Health Records (PHRs or EPRs): Electronic applications that allow patients to access, manage and share their health information [20,38].
- Telemonitoring and Web applications: Electronic systems that allow patients to remotely transfer data to one or more health care providers [17].
- Web-based resources: These may include health information websites and online peer-to-peer support groups [35,39].

Electronic PHRs and telemonitoring systems can offer opportunities for improved continuity of care, efficiency, decision-making support, and greater partnerships between patients and their caregivers and providers [38]. One qualitative study conducted by Woods et al found that patient access to their EMR information improved patient-provider communication by (1) enhancing in-person communications, (2) helping patients to remember what was said at in-person visits, (3) helping patients to prepare for future appointments, and (4) helping patients to coordinate with their other providers [40]. Accessing EMR information was also found to improve patient self-management and supported shared decision-making between patients and providers.

There have been a number of studies examining the use of eHealth technologies to monitor patients on an ongoing basis. Two recent systematic reviews found that eHealth-supported monitoring can improve outcomes for patients with chronic illnesses (including diabetes, asthma, hypertension, and cardiac obstructive pulmonary disease) [16,34]. One of the reviews also found evidence that monitoring symptoms helped patients with the self-management of their care, leading to improved health benefits, patient satisfaction, and reduction in physician visits and appointment times when compared to standard care [16]. Whether eHealth-supported monitoring will improve outcomes and self-management for CCDD patients is yet to be seen given that there are few tools designed for this population. We might expect, however, that outcomes identified above may be highly beneficial for participants in our study who identified difficulties with self-management and with accessing providers due to transportation or cost issues.

eHealth Tools to Support Provider-Provider Interactions

Participants in our study identified a number of problems associated with a lack of communication between their multiple providers. Sharing patient information, for instance through a commonly accessible EMR, was identified as an important step towards improving interprovider communication and as means to improve the continuity and coordination problems participants experienced. There have been many calls in the literature to use EHRs, EMRs, and PHRs to support integration, care coordination, and continuity [36,41-45]; however, not all electronic systems are created equal.

EMRs may be useful for intra-organizational coordination and continuity but limited when it comes to supporting interorganizational communication. A qualitative study of physician use of EMRs in the United States found that while EMRs were able to facilitate within-office care coordination, the lack of standardization and inadequate operational processes limited their capacity to encourage coordination between different health care organizations [44]. What would be more appropriate, particularly for participants in our study with CCDD resulting in their having multiple providers at different organizations, would be an EHR [36] that houses patient information at a system level rather than at a single organization. Given that CCDD patients experience social, as well as medical complexity [4], there is the added challenge of making EHR data available to social service providers outside of health care, such as social workers, who may be important care team members for patients with CCDD. The need to expand our definition of providers in the context of CCDD patients will undoubtedly raise new challenges with regard to data security and privacy. Determining which providers need access to what types of information and how that access is granted will need to be addressed.

Weighing eHealth Tradeoffs

An important finding in our study is the concern of participants regarding the adoption and use of eHealth technologies. Issues of shifting responsibilities, changing patient-provider interactions and relationships, and privacy concerns identified by participants have been noted in the eHealth literature [36,39,46,47]. However, similar to findings in our study, one study overviewing patient input into the development of a new EHR system in the United States found that the patient-perceived potential benefits of an EHR system outweighed patients' concerns regarding privacy and security [48].

In addition to the potential issues with eHealth identified in our focus groups, there have been some studies to suggest that increasing patients' access to their medical information and engaging them in monitoring could actually increase anxiety [35,40]. As CCDD patients will often experience mental health challenges [4], an impact on anxiety as a result of using the tool may be a particular concern when developing monitoring technologies. While increased patient anxiety was not raised in our focus groups, primary care providers identified this as a concern through informal discussions with the research team. In designing our tools, we will ensure that we include patient debriefs and monitoring for increased anxiety so unintended adverse events can be avoided.

Limitations

A potential limitation is that participant opinions may be shaped by their perception of what is socially acceptable, which is a limitation for most qualitative studies, particularly focus groups in which participants may feel pressure to share only opinions they feel are shared by the group. Another limitation was the small size of the focus groups; in one instance, a group contained only 2 individuals. While we were still able to maintain a meaningful and rich conversation (as noted in the methods section), more individuals in the room may have spurred additional conversation that may have elicited additional concepts that were not captured. However, reaching thematic saturation suggests no new topics were likely to arise even with additional participants. It is also possible that fewer participants in the focus group allowed for more in-depth discussion and could as such be considered a strength of the study. The use of appropriate study methodology and rigorous analysis approach is another notable strength.

One important limitation may be that many participants had noted an existing comfort with mobile and computer technologies. It is possible that a less technologically savvy group may not have been so positive about the potential for eHealth technologies to help support their needs. However, one participant refused to use the technology, and a few others were not as comfortable with the technology, and their concerns were reflected in our study.

Implications for Development of Our Tools

These study findings provide us with important groundwork to start the development of eHealth tools to support community-dwelling patients with CCDD. We are encouraged that the participants in our study perceive that eHealth technologies could be beneficial to supporting their needs in primary care settings. Our focus group participants identified that they require improved patient-primary care provider communication, improved interprovider communication, and that while eHealth technologies can offer a number of benefits, there are potential tradeoffs that researchers and developers should take into consideration. We will begin with a focus on

developing telemonitoring to support ongoing patient-provider interaction and patient self-management. As our health care system in Ontario is far from having an integrated EHR, our monitoring tools will include a portal system to allow patients to share data with multiple providers.

A key challenge we, and many others working in eHealth, face are the challenges in supporting interprovider communication. In particular are the barriers associated with creating a commonly accessible EHR such as lack of standardization of clinical information, patient concerns over security and privacy, provider concerns over legal liability, and costs [36]. Given these barriers, a first step forward may be simply providing patients with CCDD mobile access to their medical records that they could then share with their multiple providers at the point-of-care or by giving providers access to a Web-based portal. Although, we could design a tool that allows for multiple provider access, implementing this strategy in a fractured system, as is the case in Ontario and much of Canada, is likely to be a challenge. Through piloting we will determine the feasibility of this approach and identify other options for improving interprovider communication to support patients with CCDD.

In order to avoid the potential pitfalls of eHealth technologies identified by our participants, we will adopt a user-centered design approach to develop our tools, allowing us to design and implement our tools in partnership with patients. User-centered design fits within the broader design evaluation approach used for our project and supports our aim to keep patient and provider users heavily involved in the full development process. In keeping with the user-centered design methods, we anticipate multiple iterations of our tools that will be reviewed by both patients and providers at each step. While the literature identifies the potential and realized benefits of eHealth tools, many of these tools and studies focus on patients with single diseases. Our tool will be addressing a notable gap in eHealth technology through the development of patient-centered tools specifically for patients with CCDD.

Conclusions

From our patients' perspectives, there is a significant potential for eHealth tools to support patients with CCDD in community and primary care settings through enhancing two-way communication between patients and providers, and care coordination and continuity through improved interprovider communication. However, we need to be wary of the potential downfalls of adopting eHealth technologies and pay special attention to patient-identified needs and concerns. We are thus encouraged that the patient-centered eHealth tools we intend to build will be able to address the many challenges faced by patients with CCDD at our particular setting. As we move into the piloting and evaluation phases, we will seek to roll out the tool more broadly to other team-based primary care settings. The strength of our approach is in using patient-identified needs to drive tool development, allowing us to build patient-centered tools and support patient-centered care more broadly.

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Authors' Contributions

CSG was the lead author and contributed to conceptual development, data gathering and analysis, framing the paper including logic/rationale for the study, arguments to be included in all sections, and integrating co-author feedback for final submission. DM contributed to data gathering and analysis, feedback on manuscript drafts including framing and arguments. KK contributed to conceptual development, data analysis, and feedback on manuscript drafts including framing and arguments. CC contributed to conceptual development, data analysis, and feedback on manuscript drafts including framing and arguments.

Conflicts of Interest

None declared.

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Abbreviations

CCDD: complex chronic disease and disability
EHR: electronic health record
EMR: electronic medical record

FHT: family health team
PHR: personal health record

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Original Paper

Design and Usage of the HeartCycle Education and Coaching Program for Patients With Heart Failure

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Abstract

Background: Heart failure (HF) is common, and it is associated with high rates of hospital readmission and mortality. It is generally assumed that appropriate self-care can improve outcomes in patients with HF, but patient adherence to many self-care behaviors is poor.

Objective: The objective of our study was to develop and test an intervention to increase self-care in patients with HF using a novel, online, automated education and coaching program.

Methods: The online automated program was developed using a well-established, face-to-face, home-based cardiac rehabilitation approach. Education is tailored to the behaviors and knowledge of the individual patient, and the system supports patients in adopting self-care behaviors. Patients are guided through a goal-setting process that they conduct at their own pace through the support of the system, and they record their progress in an electronic diary such that the system can provide appropriate feedback. Only in challenging situations do HF nurses intervene to offer help. The program was evaluated in the HeartCycle study, a multicenter, observational trial with randomized components in which researchers investigated the ability of a third-generation telehealth system to enhance the management of patients with HF who had a recent (<60 days) admission to the hospital for symptoms or signs of HF (either new onset or recurrent) or were outpatients with persistent New York Heart Association (NYHA) functional class III/IV symptoms despite treatment with diuretic agents. The patients were enrolled from January 2012 through February 2013 at 3 hospital sites within the United Kingdom, Germany, and Spain.

Results: Of 123 patients enrolled (mean age 66 years (SD 12), 66% NYHA III, 79% men), 50 patients (41%) reported that they were not physically active, 56 patients (46%) did not follow a low-salt diet, 6 patients (5%) did not restrict their fluid intake, and 6 patients (5%) did not take their medication as prescribed. About 80% of the patients who started the coaching program for physical activity and low-salt diet became adherent by achieving their personal goals for 2 consecutive weeks. After becoming adherent, 61% continued physical activity coaching, but only 36% continued low-salt diet coaching.

Conclusions: The HeartCycle education and coaching program helped most nonadherent patients with HF to adopt recommended self-care behaviors. Automated coaching worked well for most patients who started the coaching program, and many patients who achieved their goals continued to use the program. For many patients who did not engage in the automated coaching program, their choice was appropriate rather than a failure of the program.

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KEYWORDS

e-counseling; heart failure; lifestyle; patient adherence; self-care; telehealth

Introduction

Heart failure (HF) is common and associated with high rates of disability, hospital readmission, and mortality, all of which can be improved by high-quality care [1]. However, management is complex, and health care resources are limited. Enabling and empowering patients, as well as their informal caregivers, to be active participants in the delivery of care (ie, self-care) could improve the quality of care in an efficient and affordable manner [2]. Self-care behaviors of patients with HF include taking medication as prescribed, engaging in physical activity and exercise, eating a low-salt diet, restricting fluid intake, and monitoring signs and symptoms [1]. Patients with HF who report effective self-care have lower mortality and readmission rates than those who report poor self-care [3]. Therefore, engaging patients in self-care is advantageous for both health care systems and patients themselves.

In order for patients to be able to engage in optimal self-care, they and their caregivers are educated about HF and how they may contribute to their own health care [4]. Education may take place during hospitalization, at the outpatient clinic, in the community, or at home. It is typically delivered by health care professionals in face-to-face sessions and supported by booklets or digital media (eg, CD-ROM [5], websites [6], telehealth systems [7], and tablet computers [8]).

Unfortunately, adherence to most self-care behaviors is remarkably low among patients with HF [9,10]. There are several reasons that may contribute to this. First, despite current recommendations, many hospitals fail to provide adequate education and staff with this designated responsibility [11,12]. The workload of health care professionals is high and is unlikely to decrease, owing to rising health care costs, the current economic downturn, and concomitant cutbacks in staffing [13]. Second, up to 73% of patients with HF have diminished cognitive function [1,14,15], which makes it difficult for them to understand, remember, and apply what they have been taught [16]. Third, many patients with HF have depression [17], which may interfere with their ability to learn and may reduce their motivation to change their behavior [1]. Fourth, even when people have sufficient knowledge, initiating and maintaining behavior changes can be challenging [18,19].

One of the aims of the European Union's Seventh Framework Programme HeartCycle project was to investigate how telehealth systems could be used to increase adherence to self-care behaviors among patients with HF. At the start of the HeartCycle project in 2008, we analyzed existing efforts to promote self-care among patients with HF and observed that most patients receive self-care education, but get little practical support at home to implement what they have learned. Telehealth systems were focused on the monitoring of vital signs, and, although education on self-care was incorporated in some of them [20], none included coaching patients in changing their behaviors. Researchers in several trials had studied the counseling of patients with HF via telephone interventions performed by trained nurses [21-23]. Although these telephone interventions increased adherence, their main disadvantage was that they were labor-intensive and therefore costly.

In order to have a more cost-effective approach, in the HeartCycle project we developed a novel automated education and coaching (E&C) program and implemented it in a telehealth system. The intention behind this program was not to replace contact with health care professionals, but to enhance the provision of information and to enable behavioral adaptation by means of automated coaching, allowing direct health care consultations to be targeted to more complex needs. We hypothesized that patients can be engaged in HF self-care behaviors by adding a coaching component that uses motivational prompts and feedback to increase their feelings of confidence in and perceived importance of these behaviors, as well as self-regulation tools to let patients set goals and monitor their own progress.

In this paper, we describe the design and use of the HeartCycle E&C program in a multicenter observational trial.

Methods

Overview

The HeartCycle E&C program was developed to provide patients with education on HF and associated self-care behaviors and to coach them in adopting these behaviors. In this section, we first summarize the scope, theoretical basis, and delivery infrastructure of the program and then focus on the program itself.

Program Ingredients

The 2008 guidelines for the diagnosis and treatment of HF issued by the European Society of Cardiology [24] were taken as a starting point to identify the topics and self-care behaviors to be addressed in the program. We decided to offer E&C for daily monitoring of signs and symptoms, physical activity, medication intake, low-salt diet, and fluid restriction. For other topics, such as alcohol consumption and sleep disorders, the program offered education and tips but no explicit coaching. For topics that were considered too sensitive to be dealt with via a telehealth system, such as sexual activity and prognosis, patients were encouraged to seek individual support from their health care team.

Several behavior change theories exist in health psychology, such as the trans-theoretical model of behavior change and the health belief model. Our program is based on the behavior change approach of the Heart Manual. The Heart Manual is the United Kingdom's leading home-based self-management program for individuals recovering from acute myocardial infarction (AMI). It is facilitated by trained health care professionals, and, through the combination of education, support, and behavioral adaptation, it supports patients after an AMI in many of the same lifestyle behavior changes recommended to patients with HF. The program is based on cognitive behavioral therapy and self-regulation theory, and it employs motivational interviewing. This home-based program is as effective as those administered in cardiac rehabilitation centers [25-28].

To focus the research on self-care rather than on the delivery infrastructure, we chose to reuse the existing Philips Motiva telehealth system, which enables patients to record and transmit

vital signs data (eg, weight, blood pressure, pulse) and displays text and videos via a set-top box and TV. The patient uses a remote control to navigate through menus, to select and control educational videos, and to answer multiple-choice questionnaires. The set-top box communicates with a secure back-end server via a secure Internet connection that can be accessed using a PC-based dashboard by authorized HF nurses.

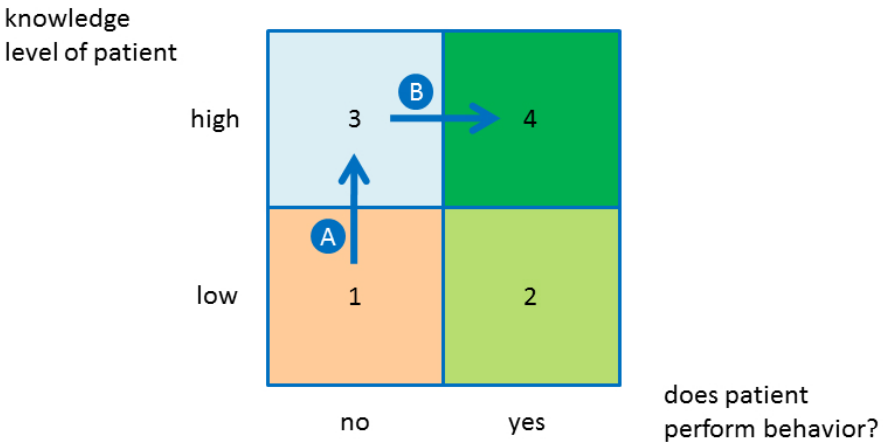
Program Description

Overview

The content presented to the patient depends on the patient's current behavior and knowledge level. An overview of the educational and behavioral possibilities is shown in Figure 1. Ideally, the patient should already have adopted the self-care behavior and know why it is important (ie, the patient is in area 4 for this behavior). If the patient knows what is supposed to be done but does not do it (area 3), the system offers coaching for this behavior (arrow B). If a patient neither knows what to do nor does it (area 1), the system first offers education to increase the patient's knowledge of this behavior (arrow A) and then provides coaching (arrow B). This mirrors the Heart Manual approach to cardiac rehabilitation, in which the facilitator seeks to first assess the patient's knowledge and then current behavior, and then provides education and support to target the areas of need.

The remainder of this section describes the steps of the program. Example screenshots are included in Multimedia Appendix 1.

Figure 1. The education and coaching framework. Arrow A denotes education, and arrow B denotes coaching.



Behavior and Knowledge Assessment

For each self-care behavior, the system determines the position of the patient in the matrix shown in Figure 1 by presenting behavior and knowledge questions to the patient. The questions are based on the validated European Heart Failure Self-Care Behaviour Scale [29] and the Dutch Heart Failure Knowledge Scale [30]. However, to assess the patient's behavior further, we introduced additional questions. For example, we added the statement "I read the label information on food packages to know their salt (sodium) content" to the scale's statement "I eat a low salt diet." Furthermore, to make it less likely for the patient to guess the correct answer, we added the answer "I don't know" to all knowledge questions.

Education

Upon completion of the behavior and knowledge assessment, the patient is given access to educational material (videos and textual tips) via the system. The videos and tips are related to the knowledge and behavior topics in the questionnaires. The videos (see Multimedia Appendix 2) contain animations, role models, and expert interviews. If a patient's score indicates insufficient knowledge of a behavior that he or she is not engaging in, the patient is prompted to watch the corresponding videos before starting with coaching. Hence, the educational content is tailored to the patient's needs, thus making it more relevant and reducing resistance to the program.

Importance and Confidence Assessment

In the motivational interviewing approach to behavior change, an individual's readiness to change determines the likelihood of attempting to change behaviors [18]. Readiness to change is related to the importance the person places on the change and how confident the person is in making the change. Therefore, for each behavior that a patient is not engaging in, the patient is asked to rate its perceived importance and how confident he or she is in adopting that behavior, and then the patient is given feedback. As low importance is often related to misconceptions [31], the system delivers messages about the most common myths associated with the behavior (eg, "Physical activity is not safe for people with heart failure") and the corresponding truths (eg, "It is important to be physically active and to rest regularly in between"). For patients with low confidence, which is often related to practical barriers in adopting the behavior [31], the system also gives them information about related solutions and tips. This feedback should address common misconceptions and raise the patient's motivation to engage with the coaching program.

Coaching

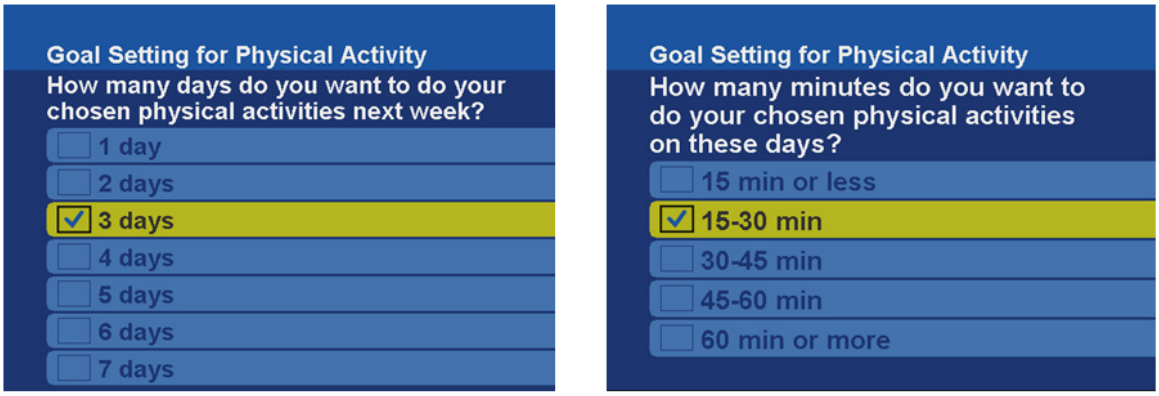
The patient starts the coaching program by setting a personal goal for the following week. The goal depends on the self-care behavior. For example, for physical activity, patients indicate the number of days they want to be active during the following

week (eg, 3 d), as well as the number of minutes they will engage in physical activity on those days (eg, 15-30 min/d) (see Figure 2). Every day, the patient enters progress data into the system by means of an on-screen diary. For example, for physical activity, the patient selects the number of minutes (eg, 15-30 min) and rates the effort of the activity (eg, too hard).

At the end of the week, the system provides the patient with feedback on progress. The exact content of the feedback message is tailored to the patient's goals, progress, and effort rating. For example, if the patient was physically active on fewer days than planned, and the sessions on those days were shorter than planned and too hard, the system gives the patient advice to set a less ambitious goal for the next week.

If the patient continually experiences difficulties in reaching personal goals, the system recommends contacting the HF nurse for further advice and support. Furthermore, it generates an alert in the HF nurse's dashboard, such that the nurse may decide to call the patient. Hence, although the routine parts of the coaching are automated, the system relies on the nurses for the most challenging parts. This is crucial because, in this way, the nurses can focus on the patients who really need their personal attention, thus allowing an efficient allocation of resources.

Figure 2. Screenshots from the telehealth system for goal setting for physical activity.



Adherence

We defined successful adherence to a self-care behavior as reaching personal goals for 2 consecutive weeks. This is a short time frame for achieving sustained behavior change. However, patients were asked to keep a daily on-screen diary, which could have become burdensome over time. We did not want to run the risk that patients would not adopt self-care behaviors because of the progress entry effort, so we chose a 2-week time frame.

When a patient has become adherent, the system acknowledges the patient's achievement and asks whether the patient wishes to continue to receive coaching. If the patient wishes to continue, the system asks the patient to set new goals for the next week. Otherwise, in 2 months, the system will ask the patient whether this behavior has been maintained. If the patient has relapsed, the system offers the possibility to receive coaching again. Hence, for patients for whom the aforementioned 2-week time frame was too short to achieve sustained behavior change, the system offers a new opportunity to receive coaching.

Eligibility and Study Design

The evaluation of the E&C program was part of the HeartCycle study. Briefly, HeartCycle was a multicenter observational trial with randomized components in which we investigated the ability of a third-generation telehealth system to enhance the management of patients with HF.

The study had four phases. Phase 1 was an observational period to familiarize the patient with the system, measure the patient's compliance with monitoring, and assess the patient's ability to achieve target doses of medications specified in an individual patient care plan, based on European Society of Cardiology guidelines. Care plans were constructed for each patient by an experienced clinician at the site, taking into account factors such as blood pressure, serum potassium concentration, and renal function to modify guideline target doses. Phase 2 comprised 2 randomized cross-over studies. One was for patients with well-controlled symptoms and signs at the end of Phase 1, and compared usual care with a diuretic minimization algorithm. The other included patients with poorly controlled symptoms and signs and compared usual care with a diuretic optimization algorithm. Phase 3 was an observational period in which instructions were given to conduct programmed activities approximately twice per week, such as skipping a medication dose, taking a heavy meal or strong coffee, taking a shower, or engaging in some exercise to see what effect these had on telemonitoring values. Finally, patients entered a longer-term follow-up phase until the last enrolled patient completed Phases 1 to 3 or discontinued monitoring.

Potential participants should have had a recent (<60 d) admission to the hospital for symptoms or signs of HF (either of new onset or recurrent) or were outpatients with persistent New York Heart Association (NYHA) functional class III/IV symptoms despite treatment with diuretics. The inclusion and exclusion criteria are shown in Textboxes 1 and 2. The protocol was reviewed and approved by the ethics committee of each participating center. All patients provided voluntary written informed consent.

It was expected that most patients would be enrolled on cardiology and medical wards during recovery from an episode of worsening HF and that the remainder would be referred from

outpatient clinics by research nurses and doctors. Patients were enrolled between January 2012 and February 2013 at three hospital sites within the United Kingdom, Germany, and Spain.

Sample size and power were calculated based on the number of patients required for Phase 2. This calculation was based on the binary endpoint of patient preference for diuretic minimization or optimization compared to standard management using McNemar's test [32]. All patients participating in the HeartCycle study were offered the E&C program. However, the exact content of the program was tailored to each patient's specific deficiencies in self-care behavior at the start of the program.

Textbox 1. Inclusion criteria for the HeartCycle study.

Inclusion criteria:

- A clinical diagnosis of heart failure
  - Cause of heart failure for any reason other than those that are rapidly reversible (see exclusion criteria)
  - May include patients with and without a low left ventricular ejection fraction or with valve disease
- Requiring treatment with at least 40mg/d of furosemide or equivalent (1mg/d of bumetanide or 10mg/d of torasemide)
- Evidence of advanced or unstable disease
  - Admission to hospital for, or complicated by, heart failure currently or within the previous 60 d
  - Outpatients with persistent NYHA III/IV symptoms
- An elevated N-terminal pro-brain natriuretic peptide value within the 3 mo prior to enrollment
  - ≥1000pg/mL if in sinus rhythm, including atrio-biventricular pacing
  - ≥2000pg/mL if not in sinus rhythm

Textbox 2. Exclusion criteria for the HeartCycle study.

Exclusion criteria:

- Unwilling to comply with the protocol (Patients should be willing and able to take daily measurements at home throughout Phase 2.)
- Rapidly reversible causes of heart failure, such as severe anemia (defined as the need for a blood transfusion), thyrotoxicosis, and/or admission with rapid (>120bpm) atrial fibrillation with good ventricular function
- Inability, in the investigators' opinion, to operate or comply with the telehealth system, even with available support from caregivers and health care volunteers if available
- Inability to communicate directly or indirectly in the local language (English in the United Kingdom, German in Germany, and Spanish in Spain)
- Persons aged <18 y and vulnerable patient groups, such as those with dementia, psychotic illness, severe intellectual disability, or cognitive dysfunction

Data Collection and Instruments

The patients received the E&C program via messages, videos, and questionnaires through the telehealth system. The time at which patients viewed these items and their answers to the questionnaires were stored automatically in the telehealth system database. By analyzing a deidentified copy of the database, we gained detailed insight into the patients' interaction with the system, including the E&C program.

Results

Overview

In this section, we first summarize the characteristics of the patients enrolled in the trial. We then show how patients progressed through the assessments and the coaching process. After that, we display the goals of the physical activity and low-salt diet that patients set for themselves. We then present the reasons why several patients did not start the coaching program after completing the behavior and knowledge assessment.

Patient Characteristics

Of 123 patients enrolled (mean age 66 (12) y, 79% men; see Table 1), 66% were in NYHA class III, indicating that they had marked limitation in physical activity.

Table 1. Baseline characteristics of the study population (N=123).

Characteristics		n (%) or mean (SD)
Men		97 (79)
Mean age in years (SD)		66.2 (11.8)
Age >70 y		49 (40)
BMI, kg/m ^{2}		
	Underweight, <18.5	2 (2)
	Normal, 18.5-25.0	37 (30)
	Overweight, 25.0-30.0	45 (37)
	Obese, ≥30.0	39 (32)
Cardiovascular history		
	Myocardial infarction	58 (47)
	Revascularization	52 (42)
	Valve surgery	11 (9)
Comorbidities		
	Cancer	16 (13)
	Diabetes	53 (43)
NYHA [a] functional class/symptoms		
	NYHA III	81 (66)
	Angina	18 (15)
	Peripheral edema	41 (33)

[a]NYHA: New York Heart Association

Patient Journeys Through the Coaching Process

Figure 3 shows the observed journeys of patients through the assessments and the coaching process. The model is explained by using physical activity as a specific example of behavior management. The number of patients who made a transition for physical activity at least once is shown next to the arrows. All 123 patients completed the behavior and knowledge assessment. On the basis of this assessment, 50 patients were considered not physically active and received the importance and confidence assessment on physical activity. Thirty-five patients (70%) completed the importance and confidence assessment and started the coaching program. Twenty-eight patients (80%) became adherent, ie, reached their physical activity goals for 2 consecutive weeks.

Eleven of the 28 patients who became adherent chose to stop and 17 continued the coaching program, although 9 of these latter patients subsequently stopped. The remaining 8 patients continued with the coaching program until the end of the study (number not shown in Figure 3), remaining in the state "continued coaching." Every time these patients achieved their personal physical activity goal for 2 consecutive weeks, they indicated that they wanted to continue the coaching program. One patient indicated this 15 times.

Every 2 months, adherent patients who declined further coaching were sent a questionnaire asking whether they were still adherent (ie, physically active). Of 13 patients, 3 indicated that they had relapsed and that they wanted to receive physical activity coaching again. One patient had relapsed but did not want to try again. Nine patients (number not shown in Figure 3) indicated that they had remained adherent until the end of the study.

For each self-care behavior, Table 2 shows the number of patients who entered a particular state per self-care behavior. For example, for physical activity, 50 (41%) of the 123 patients were nonadherent, 35 (70%) of those 50 patients started coaching, and 28 (80%) of those 35 patients became adherent. Of these latter 28 patients, 11 stopped and 17 continued with coaching after becoming adherent. The numbers for physical activity are also shown in Figure 3.

The distribution of the responses to follow-up questionnaires from patients who stopped coaching after becoming adherent is shown in Table 3. For example, for physical activity, 13 patients received the follow-up questionnaire. Of these 13 patients, 9 reported that they had maintained the behavior, 3 had relapsed and started coaching again, and 1 had relapsed but did not retry coaching.

Because the number of patients who indicated that they did not restrict their fluid intake or did not take their medications as prescribed was very low (6 each; see Table 2), these 2 self-care behaviors are not analyzed or discussed further.

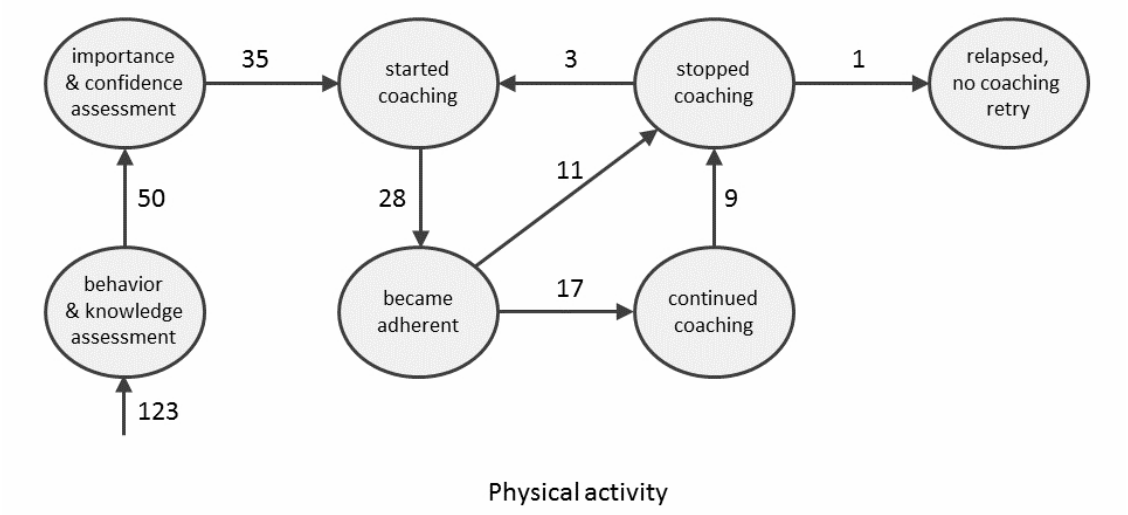
Table 2. Number of patients who entered a state per self-care behavior (N=123).

# of patients	Physical activity n (%)	Low-salt diet n (%)	Fluid restriction n (%)	Medication intake n (%)
Were nonadherent	50/123 (40.7)	56/123 (45.5)	6/123 (4.9)	6/123 (4.9)
Started coaching	35/50 (70.0)	47/56 (83.9)	4/6 (66.7)	4/6 (66.7)
Became adherent	28/35 (80.0)	36/47 (76.6)	2/4 (50.0)	4/4 (100.0)
Stopped coaching immediately	11/28 (39.3)	23/36 (63.9)	1/2 (50.0)	1/4 (25.0)
Continued coaching	17/28 (60.7)	13/36 (36.1)	1/2 (50.0)	3/4 (75.0)
Stopped coaching later on	9/17 (52.9)	6/13 (46.2)	0/1 (0.0)	1/3 (33.3)
Continued coaching until study end	8/17 (47.1)	7/13 (53.8)	1/1 (100.0)	2/3 (66.7)

Table 3. Patient responses to follow-up questionnaires (N=123).

# of patients	Physical activity	Low-salt diet	Fluid restriction	Medication intake
Received follow-up questionnaire	13	15	1	2
Maintained behavior	9 (69%)	14 (93%)	1 (100%)	2 (100%)
Relapsed and started coaching again	3 (23%)	0 (0%)	0 (0%)	0 (0%)
Relapsed but did not retry coaching	1 (8%)	1 (7%)	0 (0%)	0 (0%)

Figure 3. The states (circles) and transitions (arrows) in the coaching process. The numbers at the arrows indicate the number of patients who made this transition for physical activity at least once.



Goals When Becoming Adherent

We defined a patient as adherent to a self-care behavior if the patient reached a personal goal for 2 consecutive weeks. This section shows the goals that patients set and reached in the second of these 2 weeks.

Table 4 shows the physical activity goals (ie, combination of days and minutes) that patients reached. For example, 3 patients reached a goal consisting of 5 d/wk and 15-30 min/d. In total, 61% (17 of 28) of the patients aimed to be (and reported that they were) active on most (≥4) days of the week, and 75% (21 of 28) of the patients aimed to be (and reported that they were) active ≥15-30 min/d.

Table 5 shows the low-salt diet goals that were reached when patients became adherent. Twelve of the patients set and reported that they reached a goal of 1g/d of sodium, and 56% (20 of 36) set and reported that they reached a goal of ≤2g/d of sodium.

Table 4. Distribution of physical activity goals reached when patients became adherent (N=28).

	# of days the patient plans to be active during the week							
# of planned minutes of physical activity on these days	1 day	2 days	3 days	4 days	5 days	6 days	7 days	Subtotal
45-60 min	-	-	-	-	-	-	-	-
30-45 min	-	-	-	-	2	1	1	4
15-30 min	-	1	5	2	3	2	4	17
≤15 min	-	1	4	1	-	-	1	7
Subtotal	-	2	9	3	5	3	6	28

Table 5. Distribution of low-salt diet goals reached when patients became adherent (N=36).

Goal	# of patients
≥5 g/d	1
4 g/d	8
3 g/d	7
2 g/d	8
1 g/d	12
Total # of patients	36

Reasons for Not Proceeding to Goal Setting

Seventeen patients indicated in the behavior and knowledge assessment that they were not engaging in all self-care behaviors but they did not start the coaching program. Some did not complete the importance and confidence assessment; others did and read the feedback, but did not proceed to goal setting. The reasons for not starting the coaching program are shown in Table 6. Only 1 patient (aged 86 y) did not start coaching because of the program itself, owing to difficulties in understanding the goal-setting component.

Of the 3 patients who were too busy to use the system, 1 was very often absent from home due to employment. Of the 5 patients who had a severe physical or mental impairment, 2 had cancer and felt that they could not cope with coaching on lifestyle activities, 2 had severe HF, and 1 (aged 38 y) had difficulty coping with the HF diagnosis. Four patients who had difficulties in understanding the system had cognitive limitations and needed help from a family member to operate the system. Most technical problems were related to a slow Internet connection.

Table 6. Reasons for not starting coaching program.

Reason	n
Death	1
Patient too busy	3
Severe physical or mental impairment	5
Lack of motivation	1
Difficulties in understanding the system	4
Difficulties in understanding goal setting	1
Unknown	1
Technical problems with the system	4

Discussion

Principal Findings

This study shows that our automated coaching program was effective for most patients who started coaching. In regard to the physical activity and low-salt diet components, about 80% of the patients who engaged in them achieved their personal goals within 2 weeks.

The mean age (66±12 y) of the study participants was relatively low, and the study population was dominated by men (79%). The mean age of patients admitted to the hospital with HF in Western Europe is about 78 years. However, the mean age at admission for men and for patients with HF and a low left ventricular ejection fraction is about 5 years younger [33]. Older

patients are more likely to have cognitive dysfunction. In addition, older patients may be less willing to participate in research, thus accounting for the low numbers of elderly patients in most studies of HF.

The physical activity goals that the patients set for themselves are consistent with the most recent HF guidelines [34]. Most patients who became adherent to physical activity goals were active on ≥4 d/wk for ≥15-30 min/d. Considering that 66% of the patients in our study had NYHA III HF, this result seems positive. Patients with HF are known to have low adherence to physical activity recommendations, so additional strategies to improve adherence are required. Home-based initiatives, such as our automated coaching program, are a promising strategy for overcoming several barriers and for improving and maintaining physical function and fitness [35,36].

Our program was also successful in increasing adherence to a low-salt diet. We included this self-care behavior in our program, taking as a starting point the guidelines for the diagnosis and treatment of HF as published by the European Society of Cardiology in 2008 [24]. The newer guidelines from 2012 question the effectiveness of salt restriction in patients with HF [34], and some people argue that accepting thirst and tasteless food is very difficult. However, in a recent study, researchers showed that individualized salt and fluid restrictions can improve signs and symptoms of HF with no negative effects on thirst, appetite, or quality of life in patients with moderate to severe HF and previous signs of fluid retention [37].

Seventeen patients who indicated in the behavior and knowledge assessment that they were not engaging in all self-care behaviors did not start the coaching program. This was sometimes due to slow Internet connections, but more often was related to patient factors such as disease severity or comorbidity that rendered coaching inappropriate.

Our study also shows that there was a large variation in the percentage of patients who continued coaching after they became adherent, depending on the behavior. For physical activity, 61% (17 of 28) of patients continued coaching after becoming adherent, but only 36% (13 of 36) of patients adherent to a low-salt diet continued coaching. This may reflect the fact that once knowledge was obtained and applied, the patients knew what to do and did not feel the need for more coaching. Alternatively, patients may feel that some goals are less important or desirable than others and thus decide not to continue to adopt that change in behavior. The follow-up questionnaires showed that 93% of patients said they maintained a low-salt diet, suggesting the first of these two explanations. The percentage of patients who indicated that they maintained the behavior when they received the follow-up questionnaire also varied based on the self-care behavior. For physical activity and low-salt diet, these data were 69% (9 of 13 patients) and 93% (14 of 15 patients), respectively.

Many patients continued to use the on-screen diary when they reached their goals, suggesting either that they felt some sort of benefit, possibly as motivational support for behavior change, or that they at least found it easy to adopt. For these patients, the benefits of the automated coaching program clearly outweighed the fact that they had to manually enter their progress in a daily on-screen diary. More than 40% of the patients who continued with coaching after they became adherent continued with it until the end of the study.

One of the limitations of this study is that the program is based on patient self-reporting. In particular, whether a patient received coaching for a particular self-care behavior depended on the patient's self-reported adherence to the behavior at the start of the program. We could have integrated objective tools such as accelerometers or pedometers for physical activity, 24-hour urinary excretion (with well-known difficulties and inaccuracies) for dietary salt intake, and medication dispensers, but resources did not allow us to do so. Similarly to other studies [38], virtually all patients in our study self-reported that they were adherent to their medication prescriptions. As a consequence, we could not test the effectiveness of the medication adherence promotion part of the program.

A strength of this automated program is that it goes one step further than the simple education and information delivery model by providing coaching (ie, explicit help in adopting self-care behaviors) in the home context. Furthermore, the program is innovative in the provision of automated motivational feedback according to the levels of importance and confidence expressed by the patient. This feedback should address common patient misconceptions and increase patient motivation to engage with the coaching program. Finally, the program takes into account the dynamics of health-related behavior changes by explicitly addressing relapse. Patients who relapse are offered the opportunity to repeat the coaching program.

Since we started the development of our program in 2008, surprisingly few initiatives using automated self-care coaching for patients with HF have been begun. To the best of our knowledge, only one study (CHF-CePPORT) has aims to establish and evaluate an e-platform for behavioral counseling and education to facilitate long-term adherence to self-care among patients with HF [39]. The investigators in the TEHAF study also addressed adherence to HF self-care [40,41]; however, although their telehealth system offers personalized advice, it does not use a goal-setting approach as ours does. Researchers in another study investigated whether mobile phones could be used for vital signs measurements and symptom monitoring by patients with HF [42,43]. Although the results of that study were very positive, the system did not offer e-counseling on self-care behaviors. Hence, as far as we know, the HeartCycle study is the first in which an advanced automated coaching approach to promote self-care among patients with HF has been developed and tested.

Adherence to most self-care behaviors is poor among patients with HF [9,10]. Various initiatives have been undertaken to remedy this problem, including in-hospital education, nurse-led disease management programs, and education via digital media. In the present study, we explored an alternative approach and achieved promising results. Nevertheless, more research is needed to understand the extent to which the various components of the program contributed to the result, to investigate how self-reporting can be replaced by more objective tools, and to achieve even higher adoption percentages.

Conclusions

The HeartCycle E&C program helped patients with HF to adopt recommended self-care behaviors. Automated coaching worked very well for most patients who started coaching, and many patients who achieved their goals continued to use the coaching program.

The patients who did not engage in the automated coaching program seemed to be inappropriate candidates for this approach. If appropriate for these patients, increased face-to-face contact with an HF nurse may be required to support self-management.

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Conflicts of Interest

WS is employed by Philips. JGC has received research funding and honoraria for advice from Philips.

Multimedia Appendix 1

Example screenshots illustrating the steps in the program.

[PDF File (Adobe PDF File), 362KB - resprot_v3i4e72_app1.pdf]

Multimedia Appendix 2

A fragment from the video on physical activity.

[MP4 File (MP4 Video), 12MB - resprot_v3i4e72_app2.mp4]

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Original Paper

Toward the Development of a Lupus Interactive Navigator to Facilitate Patients and Their Health Care Providers in the Management of Lupus: Results of Web-Based Surveys

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Abstract

Background: Systemic lupus erythematosus is an inflammatory autoimmune disease associated with high morbidity and unacceptable mortality. Information and management tools are needed to help persons with lupus cope with their illness and facilitate health care providers in the delivery of care.

Objective: The objective of the study was to assess the needs and find solutions to support persons with lupus and their health care providers.

Methods: Web-based surveys were distributed across Canada to persons with lupus and their relatives (n=3119), rheumatologists (n=517), and arthritis health professionals (AHPs) (n=226) by Lupus Canada, the Canadian Rheumatology Association, and the Arthritis Health Professions Association, respectively.

Results: The survey sample comprised 665 (21.3%) persons with lupus, 98 (19.0%) rheumatologists, and 74 (32.7%) AHPs. Among the participants with lupus, 92.4% were female, the average age was 46.8 (SD 12.7) years, 79.2% were Caucasian, and 58.8% were employed. All Canadian provinces and territories were represented. The majority (43.3%) of respondents were from Ontario. Mean disease duration was 10.2 (SD 9.5) years, and 41.9% rated their global assessment as fair or poor. There was high agreement between lupus participants and health care providers regarding disease-specific information topics. All groups rated topics related to lupus, fatigue, medications, and stress as most important. Ratings differed among lupus participants and their health care providers regarding perceived helpfulness of some of the patient tools, such as the option to view test results. Needs differed for persons with lupus based on age, sex, depression, stress, and disease activity. Differences in health care provider needs were based on amount of experience in treating lupus.

Conclusions: Information and support tools needed for persons with lupus and their health care providers were identified. These results will help guide us in the development of a Web-based Lupus Interactive Navigator as an intervention tool to help persons with lupus self-manage their disease and to facilitate heath care providers in clinical management.

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KEYWORDS

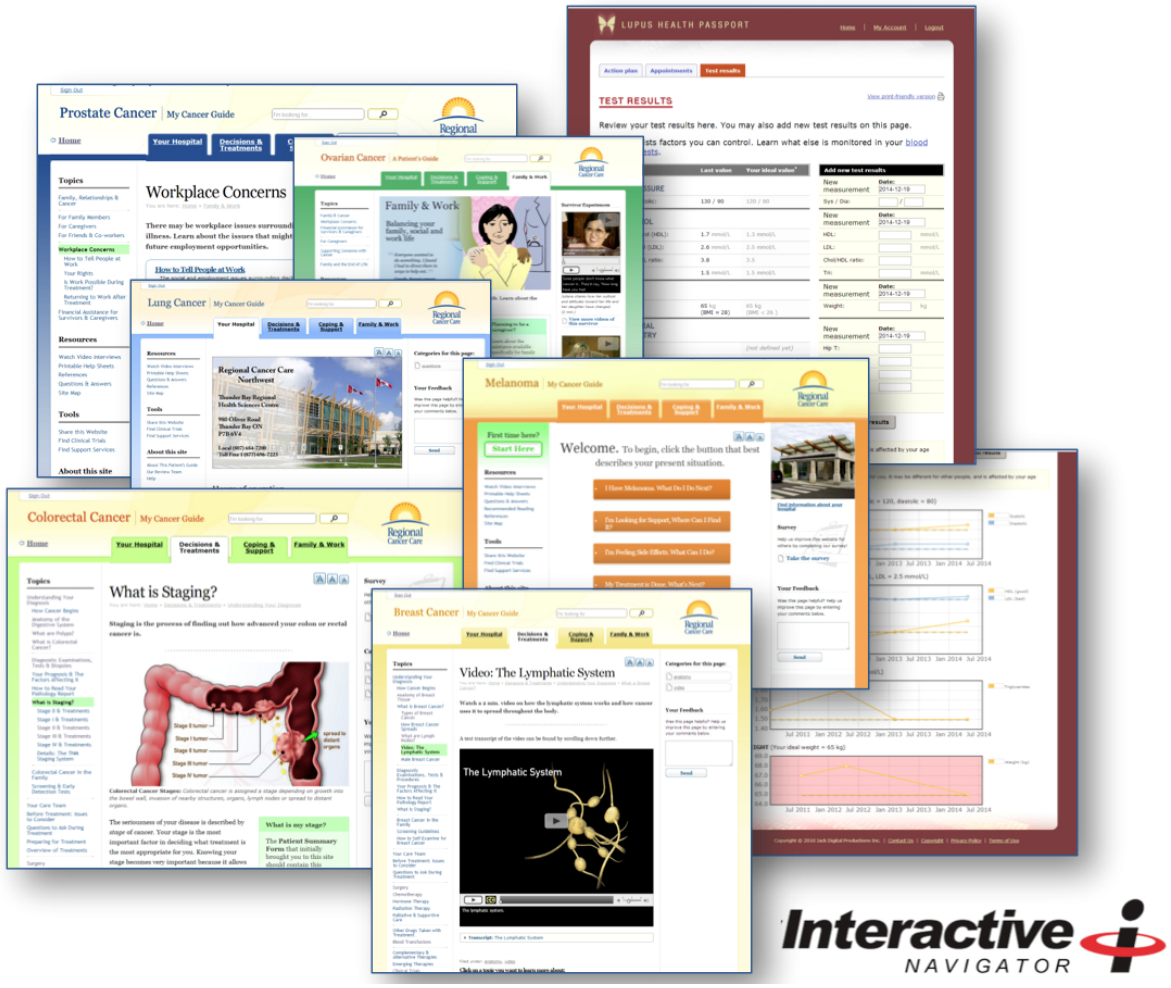
systemic lupus erythematosus; needs assessment; access to information; self-management; patient navigator

Introduction

Systemic lupus erythematosus is a chronic autoimmune disease characterized by numerous clinical manifestations. The complexity of the illness and treatment creates serious burdens to both patients and health care providers. Challenges for persons with lupus are accepting the chronic and unpredictable nature of the illness, coping with their complex care, and accessing reliable information and resources with which to manage their illness. Adapted educational and support resources for persons with lupus are limited, difficult to access, and too often irrelevant to the reality of daily living with lupus. Information and support tools are needed to help persons with lupus self-manage their disease and help facilitate the delivery of optimal integrated care by health care providers.

Recently, a successful Web-based platform, the Oncology Interactive Navigator (Figure 1), was developed to provide information and tools to help persons with cancer manage their illness and become active participants in their health care [1,2]. On the basis of these findings related to that instrument, we are initiating the development of a virtual navigation tool adapted for persons with lupus, the Lupus Interactive Navigator (LIN), to provide these individuals with up-to-date information, tailor their access to appropriate health-related resources, and help them manage and cope with their disease. To this end, we conducted Web-based surveys and distributed them to persons with lupus, rheumatologists, and arthritis health professionals (AHPs) to assess their informational needs and to guide us in the development of the LIN. In this article, we report the results of the surveys.

Figure 1. The Oncology Interactive Navigator is the prototype used for the development of the new Lupus Interactive Navigator. This figure illustrates some of the web-based pages of the Oncology Interactive Navigator.



Methods

Overview

Web-based surveys were distributed via FluidSurveys [3] to persons with lupus and their relatives (n=3119), rheumatologists (n=517), and AHPs (n=226) by Lupus Canada, which is a Canadian-based lupus patient/volunteer organization; the Canadian Rheumatology Association; and the Arthritis Health Professions Association of Canada, respectively.

Survey Development

Content

The content of the surveys was established following discussions with members of an expert panel that included a rheumatologist, a psychologist, a nurse, a person with lupus, and the developer of the Oncology Interactive Navigator. This panel had previously been responsible for the development of focus group questions used in a complementary qualitative study on this topic [4]. Through a literature review and analysis of the results of the

previous qualitative analysis, we identified informational needs of persons with lupus, rheumatologists, and AHPs, which led to our development of individualized surveys tailored for each group.

Persons With Lupus

For the persons with lupus, the surveys included questions designed to assess demographic and clinical characteristics, management strategies used, preferences regarding information topics, and tools to help manage lupus. Disease activity was assessed using a 10-point visual analog scale (VAS) (0=no activity; 10=most activity). Global assessment ratings were based on a 5-point Likert scale (1=excellent; 2=very good; 3=good; 4=fair; 5=poor).

Depression was assessed using the Patient Health Questionnaire-2 (PHQ-2) [5]. The PHQ-2 includes questions about the frequency of depressed mood and anhedonia over the previous 2 weeks and consists of 2 questions from the Patient Health Questionnaire-9 [6]. Each question is rated on a scale from 0 (not at all) to 3 (nearly every day). The PHQ-2 overall score ranges from 0 to 6. The intention of the PHQ-2 is not to establish a diagnosis, but to screen for depression. A PHQ-2 score ≥3 has a sensitivity of 83% and a specificity of 92% for detecting major depression [5]. Stress was assessed using the Perceived Stress Scale-4 (PSS-4) [7]. The PSS-4 assesses global perceived stress using 4 self-report items scored on a 4-point scale for a total possible score of 16 [8]. The response options for each item are as follows: 0=never; 1=almost never; 2=sometimes; 3=fairly often; 4=very often. The PSS-4 is not a diagnostic tool; it is used to compare stress levels within or between samples [7]. There are no established cutoffs for PSS-4. In accordance with previous work [9], we defined scores in the highest 2 quintiles as representing moderate to high stress.

Informational needs were assessed by asking participants to rate each item in a list of potential topics based on importance in managing lupus (1=least important; 10=most important). Management tool needs were assessed by asking participants to rate potential tools on how helpful they would be in disease management (1=least helpful; 10=most helpful).

Rheumatologists and Arthritis Health Professionals

Surveys for rheumatologists and AHPs included demographics (age, years in practice, and specialty), perceived barriers to providing health care for patients with lupus, and their preferences for patient information topics and tools. They were asked to rate patient information topics in terms of importance to their practice (1=least important; 10=most important) and patient management tools in terms of helpfulness to their practice (1=least helpful; 10=most helpful) in disease management. They were also asked to rate a series of clinical tools in terms of helpfulness to their practice (1=least helpful; 10=most helpful) in disease management.

All Groups

For all groups, ratings of ≥7 on individual items in the lists of information topics and tools were considered to be significantly important and helpful, respectively. Percentages were calculated based on the number of persons who rated individual items ≥7 in lists of information and tools.

Statistical Analysis

The data were transferred from Fluid Surveys [3] to Microsoft Excel v.2007 files. Means, medians, and percentages were calculated for continuous values, and percentages were calculated for categorical values.

Ethical Approval

Ethical approval to conduct the field surveys was obtained from the CHU de Québec Research Ethics Board.

Results

Persons With Lupus

Overview

The survey was mass-emailed to 3119 persons with lupus and their relatives, and we obtained a total of 808 respondents (25.90%). Of these respondents, 135 had no data related to lupus and were presumed to be relatives, and 8 were <18 years of age. We report the results of the 665 persons with lupus who responded (21.3% of original potential sample). Table 1 presents the characteristics of the persons with lupus.

All provinces and territories of Canada were represented, with the majority (43.3%) of respondents residing in Ontario. The majority (71.6%) lived in small to large urban communities, and 28.3% lived in small towns or rural communities. Among all the persons with lupus, 7.5% reported that the distance to the nearest regional hospital center was >80 km. Almost all persons with lupus (99.1%) had Web access, and most (85.5%) accessed the Web using personal computers. All respondents reported using the Web to access information about lupus, and 44.8% reported spending up to 5 h/wk for that purpose.

Table 1. Characteristics of the persons with lupus.

Characteristics	n (%)[a] or mean (SD)
Consumers and relatives contacted	3119
Consumer responders (≥18 years)	665 (21.3)
Age (n=594), mean (SD)	46.8 (12.7)
Sex, n (percent female) (n=662)	612 (92.4)
Ethnic origin, n (percent Caucasian) (n=665)	527 (79.2)
Marital status (n=664)	
Married and/or cohabiting	407 (61.3)
Single	157 (23.6)
Divorced	83 (12.5)
Widowed	17 (2.5)
Education (n=615)	
High school or less	138 (22.4)
College/university	400 (65.0)
Post-graduate/professional degree	77 (12.5)
Employment (n=660)	
Employed	388 (58.8)
Full-time	288 (43.6)
Part-time	100 (15.2)
Part-time due to SLE	60 (9.1)
Temporarily not employed	25 (3.8)
Temporarily not employed due to SLE [b]	16 (2.4)
Not employed	247 (37.4)
Not employed due to SLE[b]	175 (26.5)
Percent with access to the Web (n=650)	644 (99.1)
Work	306 (47.1)
Home	598 (92.0)
Library	58 (8.9)
Other	44 (6.8)
Device used to access the Web (n=641)	
Personal computer	548 (85.5)
iPhone or other smartphone	93 (14.5)
Web usage for information about SLE [b] **(n=641)**	
<1 h/wk	397 (61.9)
1-5 h/wk	179 (27.9)
6-10 h/wk	33 (5.1)
>10 h/wk	32 (5.0)

[a]To adjust for missing values, percentages were calculated using non-missing values

[b]SLE: Systemic Lupus Erythematosus

Clinical Characteristics

The clinical characteristics of the persons with lupus are presented in Table 2. The average disease duration was 10.2 (9.5) years. The average disease activity score reported was 4.4 (2.8). Only 3% rated their global assessment as excellent; 55% rated it as good or very good; and 42% rated it as fair or poor.

The mean depression score on the PHQ-2 was 1.9 (1.8). The proportion of persons with lupus who screened positive for depression (score ≥3) was 28.1%. PSS-4 scores revealed that almost 50% of the persons with lupus scored in the top 2 quintiles. Of these, 17.4% scored in the 4th quintile, indicating moderate levels of stress, and 30.0% scored in the 5th quintile, indicating high levels of stress.

Table 2. Clinical characteristics of participants with lupus.

		n (%)[a] or mean (SD)
Disease duration (y), mean (SD)		10.2 (9.5)
Disease activity[b] , mean (SD)		4.4 (2.8)
Global assessment (n=634), n (%)		
	Poor	58 (9.1)
	Fair	208 (32.8)
	Good	236 (37.2)
	Very good	113 (17.8)
	Excellent	19 (3.0)
PHQ-2 score (n=612), mean (SD)		1.9 (1.8)
Screened positive for depression[c] , n (%)		172 (28.1)
PSS-4 score (n=610), mean (SD)		6.9 (3.1)
Screened positive for moderate to severe stress[d] , n (%)		291 (47.7)

[a]To adjust for missing values, percentages were calculated using non-missing values

[b]Disease activity was scored on a 10-point visual analog scale (0=no activity; 10=most activity)

[c]PHQ-2 score ≥3 indicates depression

[d]Scores in 4th and 5th quintiles on PSS-4 indicate moderate and high stress, respectively; PHQ-2: Patient Health Questionnaire-2; PSS-4: Perceived Stress Scale-4 (PSS-4)

Access to Health Care

The proportion of persons with lupus who reported having a family doctor was 93.1%. The distance from home to the family doctor varied from 1 to 2400 km, with a median of 30 km (IQR 15-77.5). Travel times from home to health care providers ranged from 0.2 to 15 hours, with a median of 0.75 hours (IQR 0.5-1.3). The proportion who reported having a rheumatologist or other lupus specialist was 87.5%. Distances traveled to a rheumatologist or lupus specialist ranged from 1 to 4000 km, with a median of 70 km (IQR 36.0-150). Travel times from home to a rheumatologist or lupus specialist ranged from 0.25 to 50 hours, with a median of 1.4 hours (IQR 1-2.5).

Self-Management Strategies Used

Exercise was the most frequently used management strategy (63.7%). Yoga (21.7%) and swimming (21.1%) were reported as helpful management strategies. Other frequently used management strategies reported to be helpful included prayer (42.5%), massage therapy (33.3%), and meditation (24.1%). Several strategies reported as less available but considered to be helpful were attending self-help groups; practicing stress management; and using community services, herbal medicine, and reflexology.

Rheumatologists and Arthritis Health Professionals

Overview

Ninety-eight rheumatologists (19.0%) and 74 AHPs (32.7%) responded to the surveys. The average (SD) number of years in clinical practice reported by rheumatologists was 15.2 (12.0) (range, 6 months to 39 years). AHPs reported being in clinical practice for an average of 23.9 (12.7) years (range: 6 months to 45 years). Rheumatologists saw an average of 4.7 (4.6) patients with lupus per week (range, 0.2 to 20). Most rheumatologists (96%) reported that it would be beneficial to their practice if their patients with lupus played an active role in their own health care.

Barriers to Health Care

Rheumatologists rated patients' non-adherence to medications as the greatest barrier to health care, with 76% rating it as problematic. Patients' access to medications was also considered to be problematic (51.1%). Most problematic for AHPs were access to resources (81.9%) and patients' non-adherence to treatments (75.4%).

Information Topics

The ratings of information topics for persons with lupus, rheumatologists, and AHPs are shown in Table 3. The topics

were rated on a scale of 1 to 10 in terms of importance (1=least important; 10=most important), with percentages given for ratings ≥7.

The 3 most important information topics selected were similar across the 3 groups, with the general areas of fatigue management, understanding and coping with lupus, and medications most frequently being reported as important (84.8%-93.2%). Information about stress was rated important by slightly more persons with lupus (90.9%) and AHPs (84.7%) than rheumatologists (78.9%).

Most persons with lupus rated as important information topics related to self-management, including choosing options for living with lupus, coping with lupus, managing sleep disturbances, avoiding kidney disease, engaging in diet and exercise regimens, and managing pain (range, 80.2%-86.4%). Fewer rheumatologists rated these survey items as important (53%-66%), with the lowest percentage being for diet and exercise. Across the 3 groups, the information topics least often rated as important addressed disability insurance (48.3%-55.0%) and employment counseling (41.8%-64.4%).

Surprisingly, access to psychosocial resources was rated as important less often by persons with lupus (52.6%) than by rheumatologists (79.7%) and AHPs (86.4%). Not surprisingly, information about complementary and alternative therapies was rated as important more often by persons with lupus (68.7%) than by rheumatologists (38.5%) and AHPs (32.8%).

Table 3. Percentage reporting individual information topics as important.[a]

	Persons with lupus	Rheumatologists	AHPs
Dealing with fatigue	91.3	84.8	93.2
Understanding lupus, the disease	90.9	86.1	91.4
Understanding effects of stress on lupus	90.9	78.9	84.7
Medications used in lupus	88.7	92.4	87.9
Practical lifestyle options for living with lupus	86.4	N/A[c]	N/A
Managing sleep disturbances	85.1	N/A	N/A
Diet and exercise recommendations [b]	83.4		
Diet recommendations		53.1	58.6
Exercise recommendations		61.5	77.6
Coping with arthritis	81.3	N/A	N/A
Avoiding kidney disease	81.2	N/A	N/A
Pain management	80.2	65.8	89.8
Decision-making information	74.7	N/A	N/A
Addressing depression	73.2	69.2	89.8
Managing skin rashes	69.8	69.6	71.1
Improving communication with the health care team	68.8	64.6	66.1
Complementary and alternative methods	68.7	38.5	32.8
Knowing where to get disability insurance	55.0	50.0	48.3
Access to psychosocial resources	52.6	79.7	86.4
Employment counseling services	41.8	48.1	64.4

[a]Percentages are for ratings ≥7 in terms of importance (1=least important; 10=most important)

[b]Diet and exercise recommendations were combined in the survey for persons with lupus

[c]N/A=Not Asked

Management Tools for Persons With Lupus

The ratings of the patient management tools are shown in Table 4. Management tools for persons with lupus were rated on a scale of 1 to 10 in terms of importance (1=least important; 10=most important), with percentages given for ratings ≥7.

More persons with lupus (87.5%) than rheumatologists (59.6%) rated the helpfulness of the option to view test results in managing lupus as important. Similar numbers of persons with lupus and rheumatologists rated options to update medical information (78.5% and 70.9%, respectively) and review and update medications (75.7% and 70.9%, respectively) as helpful. Coping tools, such as journaling to record symptoms and flares and to track mood and stress, were viewed as helpful more frequently by persons with lupus (70%-72%) and AHPs (71%-74%) than by rheumatologists (39%-53%). Similar numbers of persons with lupus (65.3%) and rheumatologists

(62.0%) reported that a community resource locator would be helpful. Fewer than half of persons with lupus rated chat rooms and prednisone-tapering calendars as helpful.

Table 4. Percentage reporting individual management tools as helpful. [a]

Management tools	Persons with lupus	Rheumatologists	AHPs
Option to view test results	87.5	59.6	N/A[b]
Option to update medical information	78.5	70.9	N/A
Monitor emotional wellness	76.8	N/A	N/A
Option to review and update medications	75.7	70.9	N/A
Journal symptoms and flares	72.1	53.2	70.7
Track mood/stress levels	69.9	39.2	74.1
Community resource locator	65.3	62.0	84.5
Chat rooms	53.8	N/A	N/A
Calendars specific to prednisone tapering	41.5	N/A	N/A

[a]Percentages are for ratings ≥7 in terms of importance (1=least important; 10=most important)

[b]N/A: Not Asked

Clinical Tools

The results of rheumatologist and AHP ratings of the helpfulness of clinical tools are shown in Table 5. Among the clinical tools listed in the survey, rheumatologists most frequently rated as helpful patient reminders for screening and vaccinations (88.3%), current medication lists (85.5%), printer-friendly patient information (84.2%), and access to view medication changes made by the another physician or by the patient (84.2%). Other tools that they considered to be helpful were printer-friendly prednisone-tapering schedules (76.6%), access to tests results (73.1%), links to patients' general practitioners and specialists (72.7%), and access to anthropomorphic and clinical measures (68.8%). The fewest rheumatologists reported 36-Item Short Form Health Survey (SF-36) [10] scores (34.2%), template referral letters (49.4%), and ability to correspond with patients (55.8%) as helpful.

All AHPs rated printer-friendly patient information as helpful. Other items many AHPs rated as helpful were links to resources (93.1%), links to general practitioners and specialists (87.7%), and ability to correspond with patients (84.2%). Slightly more than half (54.4%) of them rated SF-36 scores as important.

Table 5. Percentage of rheumatologists and arthritis health professionals rating of individual clinical tools as helpful.[a]

Clinical tools	Rheumatologists	AHPs
Reminders (screening, vaccinations, etc.)	88.3	N/A[b]
List of patients' current medications	85.5	
Printer-friendly patient information (e.g., medication instructions)	84.2	100.0
Access to view medication changes made by another MD or the patient	84.2	N/A
Printer-friendly prednisone-tapering schedules	76.6	N/A
Access to tests results	73.1	N/A
Links to GPs[c] and specialists	72.7	87.7
Anthropomorphic and clinical measures	68.8	N/A
Links to resources	63.6	93.1
Access to LIN[d] to view and update medical data	61.8	N/A
SLEDAI[e] and SLICC DI[f] scores	61.0	N/A
Ability to correspond with patients to provide reminders and answer questions	55.8	84.2
Template referral letters	49.4	N/A
SF-36[g] scores	34.2	54.4

[a]Percentages are for ratings ≥7 in terms of importance (1=least important; 10=most important)

[b]N/A: Not Asked

[c]GP: General Practitioner

[d]LIN: Lupus Interactive Navigator

[e]SLEDAI: Systemic Lupus Erythematosus Disease Activity Index [11]

[f]SLICC DI: Systemic Lupus Erythematosus Collaborating Clinics Disease Index [12]

[g]SF-36: 36-Item Short Form Health Survey

Differences in Needs Based on Characteristics of Respondents

Persons With Lupus

We further evaluated whether the differences in ratings were due to respondents' characteristics.

We evaluated the responses of persons with lupus by individual characteristics to determine whether ratings differed by age; sex; disease duration; and disease activity, depression, and stress scores.

We found similar ratings among the persons with lupus aged <40 years and those aged ≥40 years regarding all information topics. However, the persons with lupus aged ≥40 years placed importance on management tools such as chat rooms and prednisone calendars less frequently than the younger participants did. Men less frequently than women placed importance on complementary and alternative therapies (47.9% versus 69.7%, respectively), journaling symptoms (56.3% versus 73.8%, respectively), chat rooms (43.8% versus 54.8%, respectively), and resource locators (55.3% versus 66.2%, respectively). Disease duration had no impact on ratings for information topics and management tools.

We found numerous differences in ratings between those with low disease activity (VAS score <5) and those with high disease activity (VAS score≥5). Participants with high disease activity more frequently reported interest in self-management topics, including depression, coping, sleep, pain, disability, psychosocial resources, and improving communication with the health care team. Also, compared to persons with low disease activity, those with high disease activity more frequently rated as important management tools such as prednisone calendars and journaling. Patients with greater lupus disease activity scored higher for depression on the PHQ-2 depression scale than those with lower lupus disease activity (37.4% versus 18.4%).

On all items, persons with lupus who screened positive for depressed mood (PHQ-2 score ≥3) were more likely than those without depressed mood to rate items as important. The greatest differences occurred with regard to topics related to depression, sleep, pain, disability, decision making, and psychosocial and community resources. Persons with lupus and depressed mood scored these items 8%-22% higher than persons with lupus who did not have depression. The greatest differences in ratings of the helpfulness of management tools were for chat rooms, tools to track mood and stress levels, and tools to monitor emotional wellness. Persons with lupus and depressed mood rated these items 8%-16% higher than those who were not depressed.

Participants with lupus who screened positive for moderate or severe stress rated most items somewhat higher (by 3%-5%) compared to those who were less stressed.

Rheumatologists

Numerous differences were observed regarding responses of rheumatologists with relatively more experience in treating patients with lupus (>5 patients per week versus ≤5 patients per week). Rheumatologists with relatively more experience rated most items higher than those who were treating fewer patients. The greatest differences were in patient self-management topics, including exercise, stress management, depression, and rash management, with more experienced rheumatologists rating these items 6%-14% higher than those treating fewer patients. Also, the more experienced rheumatologists placed more importance on patient management tools, including patient access to records to update medical information and medications. Differences between more and less experienced rheumatologists were also observed with regard to clinical management tools. Compared to rheumatologists who treated fewer patients with lupus, those with more experience gave higher ratings to clinical management tools such as access to test results, SLEDAI and SLICC DI scores, and ability to correspond with patients.

Discussion

Principal Findings

To our knowledge, this is the first large, comprehensive survey of persons with lupus and their health care providers conducted to date to identify information and tools needed to help these patients with self-management of their disease and to facilitate clinical management. The demographic and clinical characteristics of the persons with lupus were similar to the average Canadian lupus patient population and provided a wide spectrum of disease duration (0.7-19.7 years) and disease activity and global assessment ratings.

The following limitations of our study design should be noted. The surveys were distributed to persons affiliated with Lupus Canada who had Web access. The findings may not reflect the needs of persons with lupus who are not members of Lupus Canada or do not use computers. Also, although the survey was offered in both English and French, there were very few French-speaking respondents and thus, the needs of French-speaking persons with lupus were underreported.

The fact that we received responses from more than 600 persons with lupus indicates that a large number of consumers use email as a means of communication. This finding supports the feasibility of using a Web-based program to reach large numbers of persons with lupus. However, we do not know the prevalence of persons with lupus who do not have Web access; therefore, we cannot generalize our findings to all persons with lupus in Canada. Among the entire population in Canada, 83% reported having access to the Web [13].

Depressed mood and stress were present in some of the persons with lupus in our sample, with 28.0% having PSQ-2 scores suggesting depressive disorder and 49.9% with PPS-4 scores suggesting moderate to severe stress. Although the PHQ-2 is a screening tool, not a diagnostic tool, it has previously been shown to be a valid and reliable instrument [5,14] that can easily be used for research and in clinical practice owing to its brevity. The prevalence of depression in lupus is high and has been shown to be as high as 39% [15]. Our findings suggest that these brief measures may be useful in identifying those persons with lupus at high risk for depression or elevated stress and point to a need for use of an instrument such as the LIN to address mental health issues and guide the selection of resources to help manage depression and stress.

The most important information topics across all groups dealt with medications, fatigue, and management of lupus. Persons with lupus placed somewhat higher importance on disease management information, including stress and pain management, diet, and exercise, than rheumatologists did. However, the more experienced rheumatologists gave higher ratings for stress and exercise than rheumatologists who followed fewer patients. These results indicate that rheumatologists who treat relatively more patients with lupus, compared to those with less experience, are more in tune with these patients' needs.

The greatest differences in ratings observed between rheumatologists and persons with lupus were for information topics about complementary and alternative therapies and access to psychosocial resources. It is not surprising that persons with lupus placed higher importance on complementary and alternative therapies than rheumatologists did. Patients with chronic diseases have been shown to seek these therapies frequently [16]. The opinions of rheumatologists regarding use of complementary and alternative therapies may be driven by the lack of proven scientific evidence for most of them [17]. We were surprised to see that rheumatologists' ratings for information about access to psychosocial resources were higher than those of persons with lupus. These results may reflect rheumatologists' increasing awareness of the psychosocial burden on their patients and in their clinical practice. Rheumatologists have reported the lack of these resources to be one of the barriers to health care. Rheumatologists are not prepared to provide psychological help and do not have the time to address these needs. Patient access to psychosocial resources would benefit their patients and greatly relieve their clinical workload.

There were also several differences between rheumatologists and patients with regard to ratings of the helpfulness of patient tools, including options to view test results, journaling symptoms and flares, and tracking mood and stress levels, with rheumatologists rating these tools as less helpful than patients did. There remained a large difference in these ratings regardless of rheumatologists' experience levels. The ratings for the option to view test results may reflect rheumatologists' concerns that patients with lupus could become anxious and overwhelmed if not prepared with adequate information about test results and medical data. It is unclear why rheumatologists did not perceive patient options to track mood and stress and journal symptoms and flares as helpful to their practice. Perhaps the tracking and journaling options were considered to be less clinical and to be more helpful in psychological therapy than in medical practice.

The clinical tools that rheumatologists considered most beneficial to their practice were options to remind patients about screening and vaccinations, current medication lists of their patients, printer-friendly patient information, and access to view medication changes made by patients or by other physicians.

Conclusions

In this study, we identified specific informational needs and tools to help persons with lupus and their health care providers better manage lupus. Furthermore, we identified needs specific to persons with lupus based on their characteristics.

There was high agreement between persons with lupus and rheumatologists regarding disease-specific information topics. Although rheumatologists placed somewhat lower ratings of importance on topics related to information on patient lifestyle choices and self-management tools, their ratings did reflect that they felt these areas have some importance. Furthermore, rheumatologists who were more experienced in treating patients with lupus placed higher importance on some of the self-management information topics (eg, exercise, managing stress, depression) and self-management tools, including patient access to update medical information and medications. In future studies, we will focus on the topics of greatest importance to persons with lupus and their health care providers and will further tailor the LIN to the specific needs of persons with lupus based on these individuals' characteristics, including depression and level of disease activity, to best serve their needs.

Acknowledgments

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Authors' Contributions

All authors contributed to the conceptual framework of this paper as well as to the development of the content of the surveys with their own expertise as rheumatologist (PRF), psychologist (DDC), nurse (CN), and developer of the Oncology Interactive Navigator (MR). This effort was coordinated by DE. The analysis was done by CN. Interpretation of the data was done by CN and PRF. All authors contributed to, reviewed, and approved the paper.

Conflicts of Interest

Carolyn Neville, Deborah Da Costa, and Davy Eng declare that they have no conflicts of interest. Murray Rochon has a patent copyright, trademark, and marketing rights with royalties paid to JDP and is the founder of JDP, a social innovation company that is a partner organization under the PHSI grant provided by CIHR for this work. As part of the mandate of this grant, corporations are asked to partner with researchers to help effect change in health systems. This alignment and collaboration is a criterion of the CIHR grant to extend the reach and application of innovation. JDP contributes funds and expertise to develop this tool. It retains rights to content, technology, dissemination, and licensing of the tool in all jurisdictions. Paul R Fortin reports grants from the CIHR and an in-kind contribution from JDP during the conduct of this study, as well as other unrestricted funds from GSK Canada Inc, also as part of the PHSI CIHR grant partnership program.

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Abbreviations

AHPs: Arthritis health professionals
CIHR: Canadian Institute of Health Research
LIN: Lupus Interactive Navigator
JDP: Jack Digital Productions Inc
PHSI: Partnership in Health System and Improvement
PHQ-2: Patient Health Questionnaire-2
PSS-4: Perceived Stress Scale-4
SLEDAI: Systemic lupus erythematosus disease activity index
SLICC DI: Systemic Lupus International Collaborating Clinics/American College of Rheumatology Damage Index
SF-36: 36-Item Short Form Health Survey
VAS: Visual analog scale

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Original Paper

Sibanye Methods for Prevention Packages Program Project Protocol: Pilot Study of HIV Prevention Interventions for Men Who Have Sex With Men in South Africa

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Abstract

Background: Human immunodeficiency virus (HIV) prevention intervention programs and related research for men who have sex with men (MSM) in the southern African region remain limited, despite the emergence of a severe epidemic among this group. With a lack of understanding of their social and sexual lives and HIV risks, and with MSM being a hidden and stigmatized group in the region, optimized HIV prevention packages for southern African MSM are an urgent public health and research priority.

Objective: The objective of the Sibanye Health Project is to develop and evaluate a combination package of biomedical, behavioral, and community-level HIV prevention interventions and services for MSM in South Africa.

Methods: The project consists of three phases: (1) a comprehensive literature review and summary of current HIV prevention interventions (Phase I), (2) agent-based mathematical modeling of HIV transmission in southern African MSM (Phase II), and (3) formative and stigma-related qualitative research, community engagement, training on providing health care to MSM, and the pilot study (Phase III). The pilot study is a prospective one-year study of 200 men in Cape Town and Port Elizabeth, South Africa. The study will assess a package of HIV prevention services, including condom and condom-compatible lubricant choices, risk-reduction counseling, couples HIV testing and counseling, pre-exposure prophylaxis (PrEP) for eligible men, and non-occupational post-exposure prophylaxis for men with a high risk exposure. The pilot study will begin in October 2014.

Results: Preliminary results from all components but the pilot study are available. We developed a literature review database with meta-data extracted from 3800 documents from 67 countries. Modeling results indicate that regular HIV testing and promotion of condom use can significantly impact new HIV infections among South African MSM, even in the context of high coverage of early treatment of HIV-positive men and high coverage of PrEP for at-risk HIV-negative men. Formative qualitative research

consisted of 79 in-depth interviews, and six focus group discussions in Cape Town and Port Elizabeth. Analysis of these data has informed pilot study protocol development and has been documented in peer-reviewed manuscripts. Qualitative work regarding stigma faced by South African MSM resulted in finalized scales for use in the pilot study questionnaire. A total of 37 health care providers completed training designed to facilitate clinically and culturally competent care for MSM in the Eastern Cape.

Conclusions: The design of a future, larger study of the HIV prevention package will be conducted at the end of the pilot study, powered to detect efficacy of the prevention package. Data from the updated mathematical model, results of the pilot study, acceptability data, and advancements in HIV prevention sciences will be considered in developing the final proposed package and study design.

Trial Registration: ClinicalTrials.gov NCT02043015; http://clinicaltrials.gov/show/NCT02043015 (Archived by WebCite at http://www.webcitation.org/6THvp7rAj).

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KEYWORDS

HIV; prevention & control; South Africa; Truvada

Introduction

Human immunodeficiency virus (HIV) prevention intervention programs for, and research on, men who have sex with men (MSM) in the southern African region remain limited, despite the emergence of a severe epidemic among this group [1]. Visible communities of MSM continue to emerge in southern Africa, including in countries with very high generalized HIV epidemics. The understanding of these men, their social and sexual lives, and the multiple vulnerabilities and risks associated with their sexual behaviors in the African context is still in the early stages. The very existence of MSM and their networks remains a culturally sensitive issue in several sub-Saharan African countries, and same-sex behavior is illegal in most African nations [2]. Consequently, sub-Saharan African MSM remain among the most hidden and stigmatized groups at risk in the global HIV epidemic, and a group for whom optimized HIV prevention packages, appropriate for challenging local environments, are an urgent public health and research priority.

It is clear that, at present, no single biomedical or behavioral HIV prevention intervention is sufficiently effective to change the epidemic dynamics within specific risk communities or the broader population [3], and this is particularly true for the complex social and structural contexts of MSM in southern Africa. Combined biomedical and behavioral approaches offer prospects both for reducing individual behavioral HIV acquisition risks and for lowering transmission rates within communities [4,5]. Furthermore, the social and cultural stigmas surrounding same-sex behavior in southern Africa limit the safe spaces in which men can disclose their sexual behavior or seek clinically and culturally competent sexual health and HIV prevention services, reducing the reach and impact of many individual-level HIV prevention interventions.

This protocol describes a multiphase Methods for Prevention Packages Program (MP3) project. MP3s are multidisciplinary research programs funded by the National Institutes of Health that devise combination HIV prevention packages for specific populations, examine the safety and efficacy of such approaches in the target population, and conduct pilot activities to demonstrate acceptability to the target population and appropriateness and feasibility of the study design [6]. The outcomes of the MP3 are intended to inform the design of larger prevention trials powered to determine the efficacy of an intervention package. The organizations participating in the MP3 study outlined in this protocol are Emory University, Atlanta, Georgia; Johns Hopkins Bloomberg School of Public Health, Baltimore, Maryland; University of California, Los Angeles, Los Angeles, California; Desmond Tutu HIV Foundation (DTHF), Cape Town, South Africa; and Human Sciences Research Council (HSRC), Port Elizabeth, South Africa. The objective of the Sibanye (meaning “we are one” in Xhosa) Health Project is to develop and establish the feasibility of a combination package of biomedical, behavioral, and community-level HIV prevention interventions and services for MSM in South Africa, and to establish a knowledge base to inform future appropriately scaled combination prevention research and programmatic efforts.

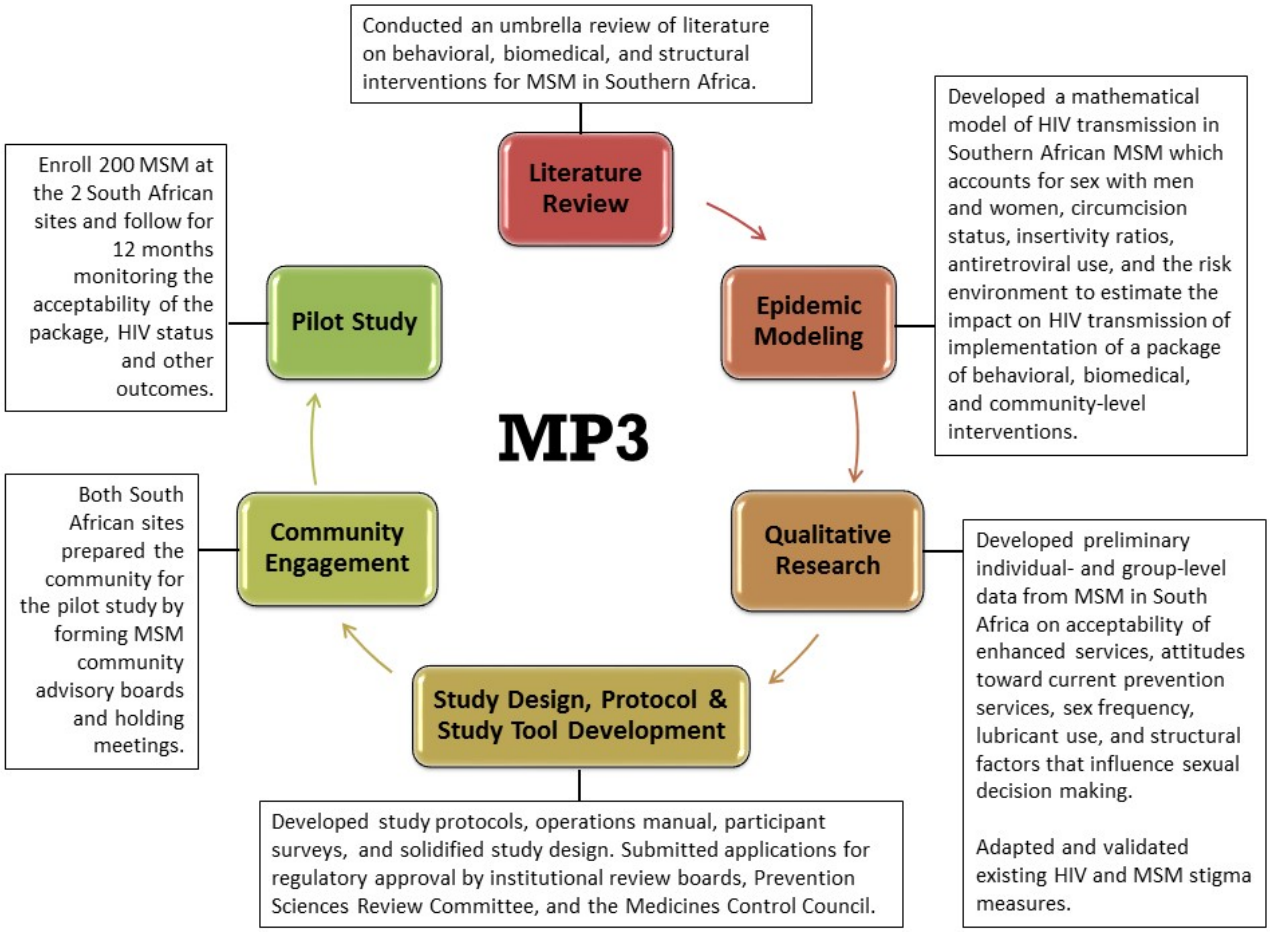
Methods

Overview

The study is being conducted in three phases (Figure 1). In Phase I of the project, a comprehensive literature review and summary of current knowledge of HIV prevention interventions was conducted resulting in the development of a database, providing inputs for the agent-based mathematical modeling effort (Phase II) and informing the components of the prevention package for the pilot study (Phase III). In Phase II, agent-based computer simulation models of HIV transmission in Southern African MSM were developed to estimate the impact that variations on a combination HIV prevention package might have on HIV transmission among MSM in southern Africa. Modeling outcomes were used to inform the components of the prevention package for the pilot study. Phase III includes both qualitative studies and a pilot study of the acceptability of the prevention package. The formative qualitative study incorporated in-depth interviews and focus groups to gain an understanding of the acceptability of intervention components and to determine optimal methods of intervention delivery. Community development, provider training, and stigma assessment were also conducted in preparation for the pilot study. The pilot study (to begin October 2014) will establish feasibility of cohort recruitment, retention, and prospective data collection of behavioral and biological outcomes in this study

setting and will provide the basis for the design of a larger prevention trial powered to demonstrate the efficacy of the combined prevention package developed through the MP3 process. Institutional review board approval was obtained by Emory University, DTHF, and HSRC and will be obtained from the National Health Laboratory Service prior to implementation of study activities. Approval from the National Institute of Allergy and Infectious Disease, Division of AIDS, Prevention Sciences Review Committee was also obtained prior to implementation.

Figure 1. Study overview.



Phase I: Literature Review

The goal of the literature review was to create a living database that could guide the development of Sibanye, and also be a public resource to other community members, researchers, and policy makers. This involved an umbrella review of HIV prevention interventions for MSM globally. There is consensus that best practices and experience from community organizations should inform development of guidelines. There is also a need to encourage dialogue between community members, program designers, and researchers to ensure that lessons learned are shared across countries and regions. Many of the strongest examples of community best practices supporting the needs of MSM in low and middle income countries remain undocumented in the sphere of peer-reviewed literature; however, these best practices are often documented in digital program reports for funders.

Therefore, we relied on several different methods to include peer-reviewed and grey literature implementation data instead of relying on a traditional systematic review. An electronic global consultation was completed in October 2011. Letters requesting information on epidemiology, rights contexts, and programming for MSM were sent out through HIV- and MSM-focused listservs in Asia, Africa, Latin America and the Caribbean, and Eastern Europe. These letters were also sent out by key funders of related initiatives including amfAR with its MSM Initiative to its grantees, the MSM Global Forum for HIV, and key United Nations agencies including the United Nations Development Programme and the Joint United Nations Programme on HIV/AIDS (UNAIDS). In addition, key informants were contacted in 28 countries requesting information specific to their country. A PubMed search (Multimedia Appendix 1) was completed to find additional peer-reviewed literature. To attain implementation data from larger implementers, Google and Bing, and the websites of large international HIV prevention implementers known to provide services for most-at-risk populations were searched with the same keywords used for the systematic searches.

We developed a database to extract the meta-data listed in Figure 2. Data extraction included author, language, title, type of author, region of the publication, and type of report including peer-reviewed studies, HIV prevention sciences work

categorized by biomedical, behavioral, structural approaches, and also non-peer-reviewed literature categorized by research, program implementation, policy analysis, guidance documents from normative agencies, and literature focused on stigma, homophobia, or homoprejudice. For documents that were freely available (ie, not peer-reviewed literature), portable document format files (PDFs) or links to the documents were included. For the remaining documents, corresponding journals were contacted to obtain rights agreements.

Figure 2. Meta-data elements extracted for materials identified in umbrella review of relevant literature, 2011-2013.

Phase II: Modeling

Agent-based computer simulation modeling was conducted to develop plausible assumptions and input parameters, inform assessment of key parameters to measure for the pilot study, and explore how different combinations of interventions to increase desired prevention outcomes could influence HIV transmission at the population level. An agent-based model was developed to simulate the impact of various combinations of HIV prevention packages. The model had a number of features including accounting for sexual networks of main, regular, and casual partners among MSM. The model used South African MSM parameters to the extent they were available based on the aforementioned literature review, and later supplemented these with qualitative data collected from the formative assessment. The endpoint was the percentage of infections that could be prevented over 5 years under a range of prevention combination scenarios [7].

HIV prevention packages that involved influencing four key prevention outcomes were examined. The four components were (1) reducing unprotected anal intercourse, (2) increasing antiretroviral therapy (ART) coverage among eligible HIV-infected persons (CD4 <350 and receipt of an HIV test), (3) Truvada-based oral pre-exposure prophylaxis (PrEP) for MSM at highest risk defined as >10 partners in 6 months or persons in sero-discordant couples, and (4) reducing the proportion of the MSM population who have never been tested for HIV by 50%.

Phase III

Formative Qualitative Assessment

Overview

To inform development of the pilot study, a series of focus group discussions and in-depth interviews was conducted with participants recruited from sites in Port Elizabeth and Cape Town, South Africa. Both qualitative methodologies were infused with a participatory learning and action approach that employed visual aids to provide structure and active participation to conversation. Detailed methods have been described previously [8]. Local languages were used in the conduct of the qualitative data collection whenever necessary.

Focus Group Discussions With MSM and Health Care Providers

Separate focus group discussions were conducted with health care providers and MSM, using semistructured interview guides (Multimedia Appendices 2 and 3). Men in the community were recruited using pre-existing contact lists of MSM interested in participating in research projects gathered by DTHF and HSRC. Once these men were screened and enrolled for a focus group discussion, further participants were recruited using snowball sampling from this initial group. Men were screened by telephone and given a brief description of the study procedures and aims. Men who were interested in participating were scheduled for a focus group discussion at one of several private locations at local venues and community-based organizations most convenient to participants. Men were selected to represent MSM from urban and peri-urban areas in Cape Town and Port

Elizabeth. Eligible participants were male at birth, reported anal sex with a man in the past 6 months, were aged 18 years or older, and spoke English, Afrikaans, or Xhosa. Health care providers were recruited for focus group discussions from local clinics and health organizations that served the communities from which MSM were recruited. All health care staff who provided any HIV counseling, testing, or treatment services were invited to participate in the focus group discussions. The primary purpose of the focus group discussions was to explore health care service experiences and priorities in clinical settings from the perspectives of both providers and MSM. Topics included clinic-based experiences with HIV prevention services for MSM, barriers to accessing or providing services for MSM, and identification of highest priority HIV prevention services for MSM. In each group, participants were shown a list of prevention options (including condoms, lubricants, and PrEP) and were asked to rank the importance of those options for MSM. For provider focus group discussions, we also assessed experience with lesbian, gay, bisexual, and transgender (LGBT) training, interest in future training opportunities, and potential formats of provider training.

Interviews With MSM

In-depth interviews were held to inform continued quantitative modeling and to inform the development of the pilot intervention package. The same methods were used to recruit, screen, and enroll men for in-depth interviews as described for the focus group discussions. Face-to-face interviews were conducted at a location convenient to the participant. The interviews included a short survey (Multimedia Appendix 4) to develop parameter estimates for mathematical modeling, such as number of sexual partners in the last 6 months and frequency of sex, by sex act, over different recall periods. Each interview used a semistructured guide (Multimedia Appendix 4) anchored in participatory learning and action approaches to explore the context of HIV prevention and risk behaviors among MSM. Participants completed a timeline activity, arranging markers for topics of interest, such as history of sexual debut, safer sex behaviors, sexual identity/outness, previous relationships, HIV testing behaviors, and involvement with community organizations (see sample timeline, Multimedia Appendix 4). Active participation was encouraged by allowing participants to select markers relevant to their lives, and discussing each in the context of other life events located on the timeline. This participatory approach sought to engage participants in the discussion and to bring a visual to make concrete the discussion of past experiences. The latter part of the interview involved a network activity, where participants listed frequented locations, sexual partners in the last 6 months, close friends, and family members. Participants were then encouraged to draw connections between elements, which facilitated conversation about how each sexual partnership was related to other components of participants' lives.

Analysis of Qualitative Data

The primary utility of the data analysis was to inform the packaging, content, and delivery of the combined prevention package. Analysis of transcribed data was guided by Grounded Theory, in particular drawing on constant comparison methodology [9] and using conceptual mapping to visualize relationships across data themes [10]. An inductive codebook was developed based on iterations of independent analysis among three coders followed by consensus revisions. Data management and analysis were conducted using MAXQDA software version 10.

Community Development and Training

DTHF has extensive experience working with MSM in the Cape Town community and has convened community advisory boards involving MSM to obtain community input on projects serving MSM. As part of community engagement activities in Port Elizabeth, the HSRC community team established a community consultation group (CCG) in July 2012 following the *Good Participatory Practice Guidelines* from UNAIDS and the AIDS Vaccine Advocacy Coalition (AVAC) [11]. This group initially consisted of key MSM stakeholder organizations such as community service, education and advocacy groups, public health representatives, and public and private sector health providers, but later became a more focused MSM CCG consisting entirely of MSM community representatives to maximize the voices of the MSM community at large. The group has more than 20 members, with approximately 10-15 members attending twice-monthly meetings to discuss a variety of health and human rights topics and receive updates on the upcoming study. The frequency of meetings has increased as the pilot study initiation nears. In essence, not only has the CCG evolved to better tailor and provide MSM community input into the study, but the CCG has become a "safe space" where MSM have been able to collectively come together from many walks of life to feel safe to discuss and collaborate on an array of topics.

Provider Training

Faculty from the Fenway Institute in Boston and HSRC conducted training in Port Elizabeth to increase the clinical and cultural competence of health care providers to provide care for MSM. A six-module training program was used to support these aims based on the *Guide to Lesbian, Gay, Bisexual and Transgender Health* developed by Fenway Health [12]. Training included information on the epidemiology of HIV and sexually transmitted infections (STIs) among MSM in sub-Saharan Africa, interacting with MSM patients including ascertaining patient sexual histories and physical sexual health examination, HIV prevention interventions for MSM, and risk reduction counseling methods. The training program consisted of didactic teaching and facilitated discussion with participants.

Stigma Measurement and Prospective Assessment

To explore how HIV and MSM stigmas compound to influence health seeking and health risk behaviors, the protocol calls for adaptation of validated stigma measures to the context of MSM in South Africa. To adapt stigma measures for MSM in South Africa, we conducted a brief literature review to identify scales that were validated for use in other populations and that fit well into Earnshaw's Stigma Measurement model [13], which focuses on anticipated, internalized, and experienced stigma. An expert panel was convened to identify preferred scales and scale items (Multimedia Appendix 5) and to recommend appropriate adaptations of items or scale formats. Following this process,

we conducted four focus groups, with 21 participants in total, to identify emic experiences with stigma for new item development. Focus groups included a pile-sorting activity to identify whether scale items aligned with Earnshaw's stigma subdomains of anticipated, internalized, and experienced stigma. This was followed by cognitive interviews to explore participant comprehension of individual scale items (Multimedia Appendix 6). Results from focus group discussions and cognitive interviews were shared with the expert review panel, allowing for consultation on the optimal tailoring and adaptation of items.

The finalized set of stigma scale items is included in the baseline questionnaire for the pilot study that will be completed by an estimated 160 HIV-uninfected respondents and 40 HIV-infected respondents. Assessment of scale validity will include determination of scale reliability and scale factors and also correlational validity based on scale correlations with theoretically supported constructs.

Analysis will assess the impact of the pilot study's provision of combination HIV prevention services on HIV and MSM stigmas. We hypothesize that for MSM enrolled in Sibanye, access to combination HIV prevention services provided in a culturally competent atmosphere will result in reductions in anticipated, internalized, and enacted MSM and HIV stigma.

Pilot Study

Overview

The pilot study will be a prospective one-year assessment of the implementation of a package of combination HIV prevention services. In addition to providing information on acceptability and uptake of the prevention package, the longitudinal study will develop capacity for conducting prospective data collection and providing prevention interventions and services. Data from the pilot study will also allow for refinement of estimates of key model parameters and support development of training curricula.

Approximately 200 MSM, 100 each in Cape Town and Port Elizabeth, will be followed for a period of 12 months. Participants will be offered prevention interventions, including condom choices with an assortment of styles, sizes, and features; condom-compatible lubricant choices, including water- and silicone-based types; couples HIV testing and counseling (CHTC) [14,15]; and PrEP for eligible men. Non-occupational post-exposure prophylaxis (nPEP) for men with an exposure at high risk for HIV transmission will be made available. Data on service utilization, condom use, HIV and STI incidence, acceptability of the prevention package, HIV-related knowledge, and other outcomes will be collected.

Participants and Enrollment

Men eligible to participate in the study will be aged 18 years and older, self-report anal intercourse with a man in the past year, be current residents of the study city, plan to stay in the city for the next year, be able to answer survey questions in English, Xhosa, or Afrikaans, be male sex at birth, be willing to provide contact information, and have a phone to facilitate the scheduling of study clinic visits. All study participants will provide written informed consent prior to participating in the pilot study.

The study will enroll both MSM who are living with HIV (HIV-positive) and those who are HIV-negative. Up to 20% of men followed for one year will be living with HIV. Additional HIV-positive men recruited will be enrolled for a baseline visit only and will not count towards the 100 men per site sample size. Recruitment and enrollment will be monitored in stages to ensure sufficient HIV-negative men are enrolled to adequately assess elements of the HIV prevention package. Enrollment of HIV-positive men will be monitored in steps. Once five HIV-positive men are enrolled prospectively at baseline, prospective enrollment of positives will pause until 20 HIV-negative men are enrolled. Men who seroconvert during follow-up will remain in the study for the full follow-up period. Recruitment activities will be conducted for approximately 3 months or until 100 MSM have been recruited and enrolled in each city. In both Cape Town and Port Elizabeth, MSM will be recruited from multiple areas including urban areas, peri-urban communities, and township areas in order to recruit diverse communities of MSM. We will employ different methods of recruitment, including event- and venue-based, online, participant referral, and walk-ins at study clinics. Men who meet the eligibility criteria during recruitment will be asked to provide contact information and be scheduled for an enrollment baseline visit.

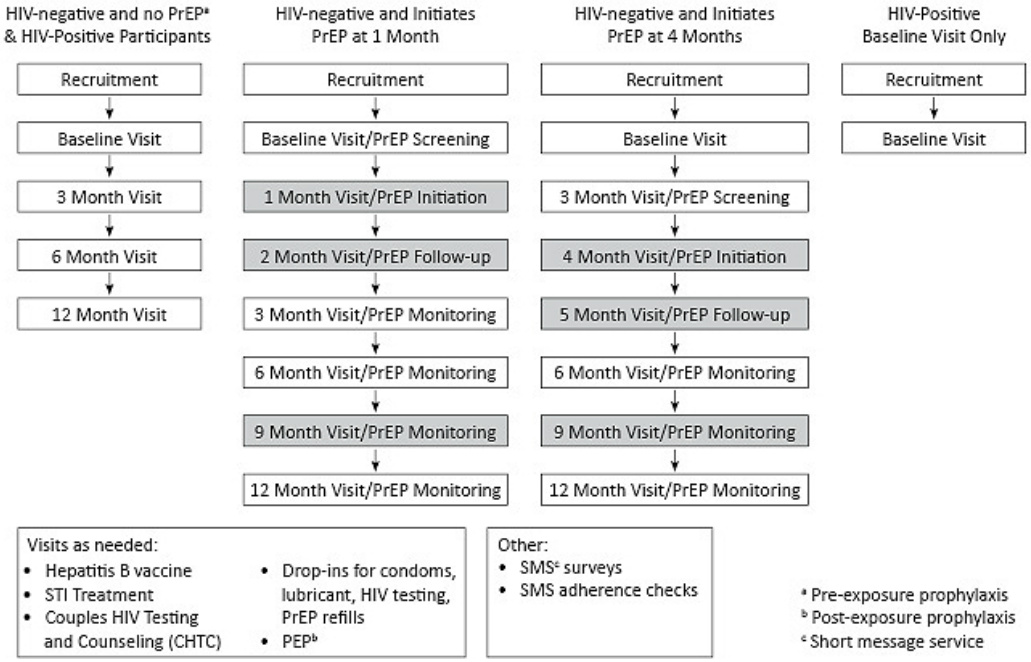
Study Design and Procedures

Men will be screened at recruitment, and eligible men will be invited to attend a baseline enrollment visit. Following consent and enrollment, the baseline visit will include a self-administered behavioral survey, HIV prevention counseling and testing, and a clinical exam (Figure 3). The clinical exam will assess STI history, circumcision status, STI and liver disease symptoms, and include laboratory testing for syphilis, urethral and rectal chlamydia and gonorrhea, hepatitis B, and urine drug screening. HIV-negative men will have additional testing for creatinine, liver enzymes (aspartate transaminase–aspartate aminotransferase/alanine transaminase–alanine aminotransferase [AST/ALT]), serum phosphate, proteinuria, and glycosuria to assess eligibility for PrEP. A dried blood spot specimen card will be prepared and stored. Men who present with STI symptoms will be prescribed medication at the visit. Participants can receive referrals for HIV treatment and care, circumcision, STI treatment (if outside the scope of study-provided treatment), alcohol and drug abuse counseling, and domestic violence counseling as needed. Men who test positive for STIs at baseline will return to the study clinic for treatment (if not prescribed medication during the visit). Men who are hepatitis B susceptible will be offered to initiate the hepatitis B vaccination series at their next study visit.

Follow-up study visits at 3, 6, and 12 months will include surveys, HIV prevention counseling, HIV testing for men who tested HIV-negative at their last visit, a clinical exam assessing STI symptoms, and blood and urine collection. At the 6-month and 12-month visits, all men will be tested for syphilis, and urethral and rectal chlamydia and gonorrhea. Men on PrEP will have additional monitoring at their standard and PrEP study

visits and additional PrEP visits 1 month after initiating PrEP, and at 9 months to assess creatinine level, AST/ALT levels, phosphorus, proteinuria, glycosuria, HIV testing, medication adherence, and to monitor side effects. Men who test positive for HIV will have blood drawn for CD4 and HIV viral load testing. Prospectively enrolled HIV-positive participants will have additional CD4 testing at 6 and 12 months and HIV viral load testing at 3, 6, and 12 months.

Figure 3. Visit schedules by participant type.



Study Intervention

The intervention package will be available starting at baseline and continuing throughout the study and will include condom and condom-compatible lubricant choices, risk reduction counseling, CHTC, linkage to care for HIV-positive men, and PrEP with emtricitabine/tenofovir disoproxil fumarate for eligible men for initiation at a 1-month or 4-month visit (Figure 4).

At their baseline visit, all participants will receive a package with condom and condom-compatible lubricant choices: ribbed, colored, flavored, fitted, thin, and flared condoms, and silicone-based and water-based lubricants. Participants will be given a Condom Scorecard to rate their condom preferences, to be returned at their next study visit (Multimedia Appendix 7). At all follow-up study visits, participants will be encouraged to pick up their preferred condom and lubricant types. Participants can also drop in to each study clinic during study clinic hours to pick up additional condoms and condom-compatible lubricant.

HIV testing will be provided to all men at their baseline visit. Risk-reduction counseling will be provided to all men regardless of their HIV status and at all standard and PrEP visits. Risk reduction counseling will be client-centered, according to the Centers for Disease Control and Prevention's (CDC) Fundamentals of HIV Prevention Counseling curriculum. HIV testing will be provided to all men at their 3, 6, and 12-month visit who tested negative at their previous visit. HIV testing will be performed by counselors trained in using South African provincial HIV testing protocols.

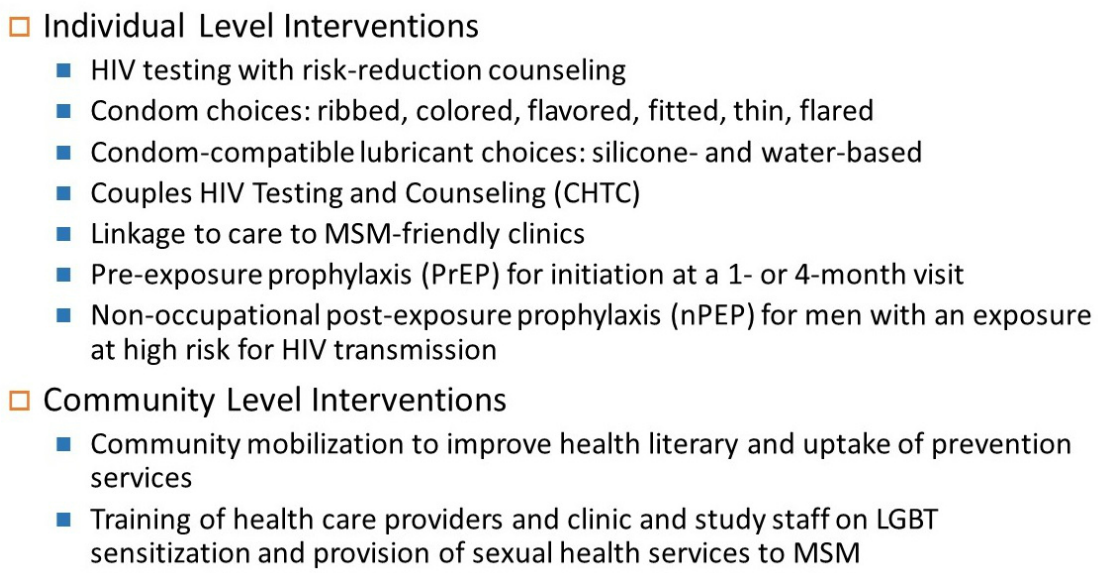
Participants will be invited to schedule CHTC appointments with a clinic counselor at any point after they complete their baseline visit. The CHTC model differs from individual testing in that a couple receives HIV testing and counseling at the level of the couple, with tailored messaging based on dyadic characteristics. Participants can attend multiple CHTC sessions with different partners; these partners do not have to be enrolled in the study to participate. All CHTC sessions will be performed by counselors who completed a 3-day training curriculum on CHTC for MSM.

Men who test positive for HIV at baseline or during the study will be linked to HIV treatment and care services as needed, including antiretroviral medication and mental health services. Referrals will be made to local providers known to be MSM-friendly.

PrEP will be available for men who meet the following eligibility criteria: HIV-negative, high-risk for acquiring HIV, adequate kidney and liver functioning, initiated hepatitis B vaccine series if susceptible, not known to have hypertension or diabetes, and willing to follow PrEP dosing and visit guidelines [16]. Men who express interest and meet eligibility criteria at their baseline visit can initiate PrEP at a 1-month initiation visit. For men who do not initiate PrEP at 1 month and later express interest and meet the eligibility criteria, PrEP will be available to initiate at a 4-month initiation visit. PrEP visits will follow the guidelines determined by the Southern African HIV Clinicians Society [16]. Throughout the study, as part of the standard of care, nPEP for men with an exposure at high risk for HIV transmission will be available.

Community-level interventions include community mobilization efforts to improve health literacy and uptake of prevention services among MSM, and training of health care providers and clinic and study staff to deliver sexual health services to MSM. Community mobilization efforts began in 2012 to build the necessary capacity to conduct the pilot study, to obtain feedback on the study protocol (eg, this feedback informed our decision to include HIV positive men), and to identify and establish appropriate and accessible clinic sites in Port Elizabeth. Community leaders and members of community organizations were consulted to determine how to include HIV prevention and health education messages as part of study recruitment events. Through engagement with the community, community members have been considered and hired in study staff positions and therefore are able to play a key role in the collection, presentation, and validation of data that characterize this population. We plan to engage with the community throughout the pilot study to further create community ownership over the interventions and facilitate uptake of the interventions. The previously mentioned provider trainings will ensure that MSM in the community have access to health care from MSM-friendly clinics and providers trained in MSM-specific health issues, improving the quality of care available to MSM in the community.

Figure 4. Intervention components.



Data Collection

Data will be collected via case report forms, self-administered surveys during study visits, and monthly short message service (SMS) surveys. Paper-based case report forms will be used to capture eligibility and enrollment data, as well as all clinical assessments, interventions, laboratory results, adverse events, and social harms reporting (Multimedia Appendix 8). All case report forms will be completed by study staff, rather than participants. These forms will be sent by scan to Emory University, with data captured using DataFax, and securely stored in participant study binders at the DTHF and HSRC offices.

At each standard study visit, participants will complete surveys on iPads or computers via SurveyGizmo, which has secure servers and a Health Insurance Portability and Accountability Act business partner agreement with Emory. Surveys will be offered in English, Xhosa, and Afrikaans (Multimedia Appendix 9). Surveys were translated from English to Xhosa and Afrikaans, then back-translated to ensure conceptual and cultural equivalence. These surveys will collect data on demographics, current use of health care services, history of HIV and STI testing, outness to health care providers, alcohol and substance abuse, history of sexual activities and condom and condom-compatible lubricant use, barriers to safer sex practices, stigma, knowledge of HIV transmission and prevention strategies, and the participant's sexual network.

An SMS survey will be sent to consenting participants each month. These brief surveys will collect information on recent sexual activity with male and female partners, including the number of partners, frequency of sex acts, condom use, lubricant use, and HIV testing outside of the study.

Outcomes

Study outcomes will be a combination of process-level outcomes, prevention impact outcomes, and acceptability and knowledge, attitudes and behaviors data, outlined in Figure 5.

Figure 5. Study outcomes.

Data Analysis Plan

The primary design for analysis will be a pre-post design. For measures that are collected from MSM participants (eg, use of condom and condom-compatible lubricant, disclosure of male sex partners to providers), data will be compared between baseline surveys and follow-up surveys. Bivariate analyses will be conducted by comparing proportions of men reporting a behavior between baseline and follow-up, using chi-square statistics. If differential retention results in follow-up data being available from a group with substantially different characteristics (eg, if older men are differentially retained, so that the 12-month participants are considerably older than the baseline sample), stratified analyses or regression modeling may be used to account for potential confounders. For biological measures, we will describe the prevalence of HIV, syphilis, chlamydia, and gonorrhea at baseline and will calculate the incidence of HIV, syphilis, chlamydia, and gonorrhea during prospective follow-up. We will conduct an intent-to-treat analysis; participants who are lost or withdraw from the study will be considered failures in analysis of uptake of the particular intervention or service of interest. HIV-positive participants who are enrolled for a baseline visit only will be included in analyses to understand prevalent behaviors and clinical features, like HIV viral load, access to care, and condom use.

Sample Size

Our pilot feasibility study is not powered to measure differences in pre-post measures in clinical outcomes; rather, we hope to develop reasonable estimates of the prevalence of certain behaviors at baseline and of the incidence of HIV infection and STIs, which may be important endpoints in a larger, future prevention trial. The choice of 100 participants per site is based on feasibility within time and budgetary constraints and will provide reasonable estimates of key parameters, including retention in the cohort and uptake of PrEP.

Results

Phase I: Literature Review

In total, 3800 documents were collected from 67 countries. Information gathered from the literature review has been made available on the Internet as a searchable electronic library. Ultimately, 2600 unique records were entered into a database, which can be viewed on the mp3docs website [17].

Phase II: Modeling

The results of the modeling activities were presented at a plenary session at the 2013 Conference on Retroviruses and Opportunistic Infections [18], and in a published manuscript [7]. The modeling results indicate that regular HIV testing and promotion of condom use produce significant and important benefits in preventing new HIV infections among South African MSM, even in the context of high coverage of early treatment of HIV-positive men and high coverage of PrEP for at-risk HIV-negative men. With a combination package that achieves a 15% reduction in unprotected anal intercourse, some PrEP

coverage, some increase in ART treatment services, and some increase in HIV testing, approximately a 35% reduction in infections can be achieved over 5 years. Modeling also found that the combined effects of the four model components to the package were not simply additive; there are some interaction effects. For example, there is an interaction between the HIV testing component and the PrEP component. If testing is increased, then PrEP as an intervention becomes more powerful because more persons become eligible for PrEP. In addition to estimating the impact of these interventions on the epidemic among South African MSM, these findings suggest that the demonstration of acceptability of a packaged prevention approach is a critical next step in optimizing prevention services for MSM in Africa.

A by-product of our modeling and simulation work was an evaluation of the sources of variation in the spread of HIV that arise from overlapping sexual networks and heterogeneity in biological and behavioral risk factors. These sources of variation are not routinely accounted for in the design of HIV prevention trials. Our work on agent-based modeling in the Sibanye MP3 project has led to useful methods for calculating required sample sizes in the design of HIV prevention trials [7].

Phase III

Formative Qualitative Assessment

A qualitative assessment involving in-depth interviews and focus groups with MSM and HIV service providers was conducted in 2012 to obtain information regarding the acceptability and optimal methods of providing HIV prevention interventions for MSM.

Findings from the qualitative data collection were used to inform the components of the prevention package. Data collection for the formative assessment, including 79 in-depth interviews and surveys, six focus group discussions with health care providers and four with MSM, was completed in early 2012. Analysis of qualitative data has informed development of the pilot study interventions. For instance, in focus group discussions participants identified access to condom-compatible lubricant as a high priority service. During in-depth interviews, a number of men discussed using petroleum-based lubricant, and these men often experienced condom breakage. In part based on these findings, we decided to include in the intervention silicone-based lubricant in addition to water-based lubricant to enhance lubricant choice and fulfill the demand for lubricant provision. The analysis of qualitative data to date has also led to a manuscript regarding the contexts of condom use [8] and a manuscript regarding repeat HIV testing [19].

Provider Training

A total of 37 providers attended the 2-day training in Port Elizabeth, including physicians, nurses, and clinical officers from across the Eastern Cape. Participants prepared for the training by reading distributed materials in advance of participation. While the goal of the training was to increase provider skills for Sibanye, a secondary benefit was the increased clinical and cultural competence of these providers in addressing the health-related needs of MSM. Among participants who were trained and completed evaluations, many of the attendees (76%, 28/37) reported that they lacked knowledge to care for MSM prior to the training. The training was well received with 100% endorsing a statement that it is important to be sensitized to MSM needs and to learn about MSM and 100% indicating they would recommend this training to colleagues.

Stigma Measurement and Prospective Assessment

An expert panel came to consensus on scale preferences and revisions to individual items for a South African stigma scale for MSM. For example, to assess the anticipated MSM stigma domain of Earnshaw's model, experts overall preferred the Liu public homosexual stigma subscale to the Pinel Stigma Consciousness Questionnaire due to use of less complex language and less focus on sexual identity. Experts also provided suggestions on individual items, including the need to allow for men of diverse sexual identities to feel included. To this end, we added an item that allows men to choose their preferred term (such as "MSM", "gay", or "bisexual"), which will automatically populate where relevant for all stigma items on the electronic questionnaire. Expert panel findings were compiled, resulting in a final set of scale items that were used for focus group discussions and cognitive interviews. Data from focus group discussions and cognitive interviews are currently being analyzed.

Discussion

Preliminary results from all components but the pilot study are available, and several products have been developed through our preliminary work. A publically accessible searchable database is now available. The findings from the modeling indicating that regular HIV testing and promotion of condom use can significantly impact new HIV infections among South African MSM have been published in a peer-reviewed journal. In-depth interview and focus group discussion data were used to inform the pilot study protocol development and will soon be published. Stigma qualitative work resulted in finalized scales for use in the pilot study questionnaire. Finally, a total of 37 health care providers in the Eastern Cape completed training on LGBT sensitization and the provision of care to MSM.

Reducing new HIV transmission by expanded, early, and consistent use of ART is key to ending the HIV epidemic [20,21]. Testing, linkage to care, and treatment in South Africa falls short of that needed to significantly decrease new infections: national data from 2012 indicate that 52.6% of women and 37.5% of men had been tested and were aware of their status [22]. Although South Africa has the largest ART program in the world [23], less than 40% of ART eligible patients are on treatment [24]. MSM also face high levels of stigma and discrimination and experience barriers to accessing health care [25]. To address these issues, other MSM-targeted programs aimed at early identification of HIV infection, decreasing HIV and STI disease burden, and improving the general health and well-being of MSM are also being evaluated. A CDC-funded implementation science project focused on MSM proposes to study community- and peer-based interventions focused on finding people who are unaware of their HIV status, increasing the linkage to care and treatment

initiation for those eligible, and achieving viral suppression through adherence support. This project will be conducted around the same time as Sibanye, comprehensively addressing multiple levels of HIV risk at different stages of the HIV treatment cascade for MSM in Cape Town, Port Elizabeth, and other areas across South Africa.

Although the goal of the Sibanye Health Project is to prevent HIV infection among MSM, the project activities align with other efforts in South Africa to identify and test persons at risk for HIV infection and optimize the continuum of HIV care for those living with HIV, which is a crucial component of comprehensive HIV prevention, treatment, and care programs. The Sibanye study activities allow men who test positive for HIV to be linked to MSM-friendly providers and services.

The design of a future, larger study of the HIV prevention package will be conducted at the end of the pilot study, powered to detect efficacy of the prevention package. The elements of the final proposed package will be determined by the study's scientific advisory board and study investigators. Data from the updated mathematical model, results of pilot studies, and acceptability data will all be considered in developing the final proposed package, and the study design. We will also take into account advancements in HIV prevention sciences that may take place during our study. For example, it is likely that additional data on the real world effectiveness of PrEP as well as efficacy of intermittent PrEP will be reported during the period of our study. Depending on the specific elements that are proposed in our final prevention package, we anticipate proposing either a neighborhood- or clinic-randomized design, or an individually randomized design. This may also be influenced by whether there are more definitive data about efficacy of specific individual-level interventions including, but not limited to, circumcision, PrEP, and nPEP.

Acknowledgments

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Conflicts of Interest

None declared.

Multimedia Appendix 1

PubMed search strategy.

[PDF File (Adobe PDF File), 34KB - resprot_v3i4e55_app1.pdf]

Multimedia Appendix 2

Focus group discussion guide - health care workers.

[PDF File (Adobe PDF File), 66KB - resprot_v3i4e55_app2.pdf]

Multimedia Appendix 3

Focus group discussion guide - men who have sex with men.

[JPG File, 102KB - resprot_v3i4e55_app3.jpg]

Multimedia Appendix 4

In-depth interview guide.

[PDF File (Adobe PDF File), 777KB - resprot_v3i4e55_app4.pdf]

Multimedia Appendix 5

Expert review scales.

[PDF File (Adobe PDF File), 100KB - resprot_v3i4e55_app5.pdf]

Multimedia Appendix 6

Cognitive interview guide.

[PDF File (Adobe PDF File), 112KB - resprot_v3i4e55_app6.pdf]

Multimedia Appendix 7

Condom scorecard.

[PDF File (Adobe PDF File), 236KB - resprot_v3i4e55_app7.pdf]

Multimedia Appendix 8

Case report forms.

[PDF File (Adobe PDF File), 1MB - resprot_v3i4e55_app8.pdf]

Multimedia Appendix 9

Baseline survey.

[PDF File (Adobe PDF File), 397KB - resprot_v3i4e55_app9.pdf]

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Abbreviations

ART: antiretroviral therapy
AST/ALT: aspartate transaminase–aspartate aminotransferase/alanine transaminase–alanine aminotransferase
CCG: community consultation group
CDC: Centers for Disease Control and Prevention
CHTC: couples HIV testing and counseling
DTHF: Desmond Tutu HIV Foundation
HIV: human immunodeficiency virus
HSRC: Human Sciences Research Council
LGBT: lesbian, gay, bisexual, and transgender
MSM: men who have sex with men
MP3: Methods for Prevention Packages Program
nPEP : non-occupational post-exposure prophylaxis
PrEP: pre-exposure prophylaxis
SMS: short message service
STI: sexually transmitted infection

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Protocol

A Phase II Multicenter Trial With Rivaroxaban in the Treatment of Livedoid Vasculopathy Assessing Pain on a Visual Analog Scale

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Abstract

Background: Livedoid vasculopathy is an orphan skin disease characterized by recurrent thrombosis of the cutaneous microcirculation. It manifests itself almost exclusively in the ankles, the back of the feet, and the distal part of the lower legs. Because of the vascular occlusion, patients suffer from intense local ischemic pain. Incidence of livedoid vasculopathy is estimated to be around 1:100,000. There are currently no approved treatments for livedoid vasculopathy, making off-label therapy the only option. In Europe, thromboprophylactic treatment with low-molecular-weight heparins has become widely accepted.

Objective: The aim of this trial is the statistical verification of the therapeutic effects of the anticoagulant rivaroxaban in patients suffering from livedoid vasculopathy.

Methods: We performed a therapeutic phase IIa trial designed as a prospective, one-armed, multicenter, interventional series of cases with a calculated sample size of 20 patients. The primary outcome is the assessment of local pain on the visual analog scale (VAS) as an intraindividual difference of 2 values between baseline and 12 weeks.

Results: Enrollment started in December 2012 and was still open at the date of submission. The study is expected to finish in November 2014.

Conclusions: Livedoid vasculopathy is associated with increased thrombophilia in the cutaneous microcirculation and the continuous use of anticoagulants helps improve the symptoms. The causes of cutaneous infarctions are heterogenous, but ultimately follow the known mechanisms of the coagulation cascade. Rivaroxaban affects the coagulation cascade and inhibits the factor Xa–dependent conversion of prothrombin to thrombin, thereby considerably reducing the risk of thrombosis.

Trial Registration: Trial Registration EudraCT Number: 2012-000108-13-DE; https://www.clinicaltrialsregister.eu/ctr-search/search?query=eudract_number:2012-000108-13 (Archived by WebCite at http://www.webcitation.org/6UCktWVCA); German Clinical Trials Register (DRKS): DRKS00004652; https://drks-neu.uniklinik-freiburg.de/drks_web/navigate.do?navigationId=trial.HTML&TRIAL_ID=DRKS00004652 (Archived by WebCite at http://www.webcitation.org/6UCIAKyCS).

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KEYWORDS

rare disease; rivaroxaban; vasculitis; leg ulcer; skin infarction

Introduction

Symptomatology

Livedoid vasculopathy is an orphan skin disease characterized by recurrent thrombosis of the cutaneous microcirculation [1-6]. The disease manifests itself almost exclusively in the ankles, the back of the feet, and the distal part of the lower legs [7-9]. Symptoms above the knee joints are rarely observed. An affection of viscera by this coagulation disorder has not been reported. Livedoid vasculopathy is difficult to diagnose due to a lack of pathognomonic markers [10-15].

Recurrent thrombus formation in the cutaneous microcirculation induces local skin necrosis and ulcerations (cutaneous infarctions) leading to irreversible atrophic scars (so-called atrophie blanche). Each cutaneous infarction inevitably leaves behind residual scar tissue, causing severe and continuous cutaneous damage in the lower extremities as the disease progresses. Typical clinical symptoms of livedoid vasculopathy are acute ulcerations and white scars (atrophie blanche), both of which appear concomitantly. In addition, the impaired blood flow caused by the disease and the accompanying reduction in oxygen saturation in the dermal microcapillaries causes a visible darkening of the skin and a vascular pattern corresponding to livedo racemosa [4,16].

Due to the vascular occlusion, patients suffer intense local ischemic pain (angina cutis). Furthermore, the acute ulcerations (cutaneous infarctions) and subsequent scarring in the ankle region lead to disfigurement of the foot and a severe reduction in patients' quality of life.

Epidemiology

Incidence of livedoid vasculopathy is estimated at around 1:100,000 [3,6]. The disease primarily affects young women, but it has also been observed in children. However, there is a significant lack of accurate epidemiological data and related patient registers. Livedoid vasculopathy fluctuates seasonally, with an exacerbation of symptoms in summer and temporary remission in winter. It is relevant to note that the estimated incidence includes patients with livedoid vasculopathy in the context of a vasculitis [17]. Based on diagnostic data from the clinics involved, it is possible to state that incidence of the prothrombotic (noninflammatory) vasculopathy described here is in fact lower than 1:100,000.

Pathophysiology

The histological examination of skin samples in livedoid vasculopathy cases shows occlusive fibrinoid material on the vascular walls and in the lumen of dermal microcapillaries [1,2,7,8,11]. Unlike vasculitis, inflammatory leukocyte infiltration is not normally detected [4,5,18,19]. Blood vessel occlusion occurs primarily in the microcapillaries of the middle and upper dermis, which are essential for blood supply to the epidermis. Occlusion of these blood vessels leads to insufficient perfusion and supply of the dependent skin area and triggers a strong pain signal. Impaired blood blow (ischemia) leads to so-called angina cutis, which if untreated causes a cutaneous infarction [7]. Tissue death caused by the absence of blood flow is then visible as necrosis and ulceration of the foot skin. The exact physiopathological mechanism of livedoid vasculopathy remains unclear. However, analysis in specific cases of the increased blood coagulation and thrombosis associated with the disease has enabled the detection of known prothrombotic markers that could help explain the hypercoagulability. This has made it possible to observe a relation between livedoid vasculopathy and several prothrombotic coagulation defects, such as factor V Leiden gene mutation, protein C or protein S deficiency, prothrombin G20210A gene mutation, anticardiolipin antibodies, hyperhomocysteinemia, or antithrombin III deficiency [4,6,20-25]. Alterations of the fibrinolytic system have also been detected in livedoid vasculopathy patients, including increased plasminogen activator inhibitor-1 and/or lipoprotein(a) levels [13,14]. Although the aforementioned markers have been detected in cases of systemic hypercoagulability, it remains unclear why livedoid vasculopathy only affects the lower extremities and is not detected in any organs other than the skin. Furthermore, it is not possible to detect pathological coagulation parameters in all patients suffering from the disease; therefore, it is expected that additional, hitherto undetected mechanisms causing thrombus formation may exist [17,23].

Treatment Options

There are currently no approved treatments for livedoid vasculopathy making off-label therapy the only option. Off-label treatment of livedoid vasculopathy seeks to prevent cutaneous microvascular occlusion, yet physicians treating the disease with anticoagulants are forced to rely on case reports and treatment series [13,20,26-30]. To date, the largest number of patients who have undergone documented treatment is 9 [31]. These patients were treated with immunoglobulins, which are nonspecific and multimodal. Immunoglobulins have an anti-inflammatory and immunomodulating effect, and it is assumed that they positively influence antibody-mediated activation of the coagulation system. This form of treatment has only been carried out at a reduced number of clinics and has not become standard practice in the treatment of livedoid vasculopathy [31,32].

Although the causes of thrombus formation are extremely varied, antithrombotic treatment is generally considered a promising area in the case of livedoid vasculopathy. However, it must be applied in the ischemic phase of the disease because the necrosis that follows this critical stage inevitably leads to irreversible scarring. In Europe, thromboprophylactic treatment with low-molecular-weight heparins (eg, enoxaparin 1-2 mg/kg body weight) has become widely accepted [28,33]. These drugs have been approved for the prevention and treatment of deep vein thrombosis. Coumarins have also been described in the literature as being helpful, and the successful use of fibrinolytics, rheological drugs, and immunoglobulins have also been reported by individual clinics [20,24].

Therapeutic Benefits of Rivaroxaban

Rivaroxaban is a novel factor Xa inhibitor used for prophylaxis of thrombosis. Treatment with the investigational medicinal product (IMP) is expected to reduce pain, prevent disfiguring after-effects, and improve the general quality of life of those treated. Experience has proven that oral administration is clearly

preferable to long-term treatment with daily subcutaneous injections of heparin necessary until now. In addition, although there is also an oral version of vitamin K antagonist Marcumar available, it requires constant international normalized ratio (INR) testing [20].

At the moment, only off-label treatments exist for livedoid vasculopathy. This means that physicians have to base their recommendations on reports of individual cases when treating livedoid vasculopathy patients. This study will provide physicians treating livedoid vasculopathy with empirical evidence for choosing the right therapy and set the basis for a possible indication expansion and inclusion in reimbursement lists.

At present, there is a delay of 5 years between the first appearance of symptoms and correct diagnosis of livedoid vasculopathy [4]. The results of this study may help other health professionals recognize and treat livedoid vasculopathy at an earlier stage. In addition, publication in scientific journals and presentation at congresses will stimulate scientific debate regarding the disease and possibly serve as a starting point for other studies.

Therapeutic Risks of Rivaroxaban

The safety of rivaroxaban was tested in 8 phase III trials with a total of 16,041 patients who received the drug by the date of protocol submission [34].

Because of its pharmacological mechanism, use of rivaroxaban may be associated with an increased risk of occult or visible bleeding in any tissue or organ, which can lead to acute posthemorrhagic anemia. Signs, symptoms, and severity (including death) vary according to location and degree of bleeding or anemia.

In clinical trials, mucosal bleeding (eg, nosebleeds, gingival, gastrointestinal, and urogenital bleeding) and anemia were observed more frequently with rivaroxaban than with vitamin K antagonist treatment [34]. Therefore, monitoring of hemoglobin/hematocrit, in addition to adequate clinical observation, may be useful in detecting occult bleeding in certain cases.

The risk of hemorrhage is higher in certain patient groups, such as those with high uncontrolled hypertension. In addition, menstrual bleeding can increase in intensity and/or duration. Some complications associated with hemorrhage are general weakness, paleness, vertigo, headache, or inexplicable swelling, as well as dyspnea and sudden shock. In some cases, an anemia can cause symptoms of cardiac ischemia, such as chest pain (angina pectoris).

Known complications of severe bleeding, such as compartment syndrome and kidney failure due to hypoperfusion, have also been reported in patients receiving rivaroxaban. Therefore, it is important that patients being treated with anticoagulants be fully aware of the increased risk of hemorrhage associated with the treatment.

Reported adverse drug reactions related to rivaroxaban have been classified according to system organ class as per the Medical Dictionary for Regulatory Activities (MedDRA) and frequency in the summary of product characteristics.

Disadvantages of Participating in the Study

The study-related disadvantages for participants are uncertainty regarding treatment success, additional time required for extra check-ups, additional time for surveys and tests, the need to register information daily in a patient diary, and answering of questionnaires.

Study Aim

The aim of this trial is the statistical verification of the therapeutic effects of rivaroxaban in patients suffering from livedoid vasculopathy. In addition to effectiveness, the study will also take into account quality of life, patient safety, and the use of rescue medications.

Methods

Trial Design

This therapeutic phase IIa (proof of concept) trial is designed as a prospective, one-armed, multicenter, interventional series of cases without a statistical interim analysis and with a calculated sample size of 20 patients (2012-000108-13-DE; DRKS00004652).

Primary Outcome

The primary outcome is the assessment of local pain on a visual analog scale (VAS): intraindividual difference of 2 values between baseline and 12 weeks.

Secondary Outcome

In addition to the primary study goal, we checked for the following secondary outcomes:

1. Assessment of local pain on the VAS: intraindividual difference of values between baseline and 4 weeks.
2. Assessment of local pain on the VAS: intraindividual difference of values between baseline and 8 weeks.
3. Assessment of local pain on the VAS: intraindividual difference of average values over 2 weeks between the first and the last 2 weeks of the treatment.
4. Assessment via the Dermatology Life Quality Index (DLQI): intraindividual difference of values between baseline and 4 weeks.
5. Assessment (DLQI): intraindividual difference of values between baseline and 8 weeks.
6. Assessment (DLQI): intraindividual difference of values between baseline and 12 weeks.
7. Consumption of rescue medication: averaged over the week, listed for every week of treatment.
8. Consumption of rescue medication: intraindividual difference of averaged values over 2 weeks between the first and the last 2 weeks of the treatment.

Statistics

Statistical analysis will be performed with descriptive methods (eg, frequency tables) and statistical parameters (eg, mean, standard deviation, and quantile). Box-and-whisker plots will be created for qualitative data and bar charts for quantitative

data as graphical methods. Moreover, inferential analyses will be carried out using appropriate significance tests and confidence intervals. Missing values will not be replaced.

For the primary outcome, the following 2-sided test problem is established: H0: $\mu=0$ versus H1: $\mu\neq0$, where μ denotes the mean of the intraindividual difference of values of the VAS for assessing local pain between start of treatment and after 12 weeks.

The primary statistical analysis will be performed with a 2-sided exact Wilcoxon test for a significance level $\alpha=.05$.

The aim of the trial is to demonstrate the therapeutic effectiveness of the study drug. Therefore, the sample size is based on this primary outcome (ie, the intraindividual difference, ["before" and "after"] in values on the VAS for assessing local pain). Therapeutic effectiveness is considered clinically relevant with a mean in the primary endpoint of at least $\Delta/\sigma=0.7$. A minimum sample size of 20 evaluable patients is necessary to demonstrate a significant therapeutic effect in the primary statistical analysis with a power of 80%.

Statistical analysis of primary and secondary endpoints will be conducted according to the intention-to-treat (ITT) principle. The ITT patient population includes all patients enrolled regardless of possible protocol deviations/violations (eg, premature termination of the study or discontinuation of study medication). In addition to ITT analysis, sensitivity analyses will be conducted according to the per-protocol (PP) principle. Relevant protocol deviations leading to exclusion from the PP analysis set will be defined in the statistical analysis plan. Definition of the analysis sets will be determined in a blinded review process without knowledge of the study endpoints.

Trial Population

It is assumed that all patients will have received some form of previous treatment for livedoid vasculopathy. For this reason, washout phases prior to the start of study treatment are defined in the inclusion and exclusion criteria.

Inclusion criteria preclude patients' participation in other clinical studies during or within 30 days before inclusion in the present study. Study participants will be informed verbally regarding possible unforeseeable health risks and possible importation of bias into the study.

At the screening visit, patients will be asked about possible associations with the investigators or sponsor that might constitute a conflict of interest relationship of dependence (eg, relatives, employees). If a relationship of dependence is suspected, the patient cannot be included in the study.

Individuals described in § 40 Abs 4 und § 41 Abs 2 and 3 of the German drug law (Arzneimittelgesetz, AMG) are excluded from participation in the study.

Men and women will be included in this study. The expected ratio of male/female patients will be 1:3. No selection according to gender will take place for study inclusion.

Inclusion Criteria

The following inclusion and exclusion criteria were defined:

1. Definite diagnosis of livedoid vasculopathy;
2. Age ≥18 and <80 years;
3. 40 points on the pain VAS on at least 1 of the 7 days prior to treatment start;
4. No participation in another intervention study within 30 days prior to treatment start;
5. Adequate communication skills in the German language; and
6. Patient must be able to recognize the nature, significance, and scope of the clinical trial and act accordingly.

Exclusion Criteria

1. Known allergy to the trial medication;
2. Known problems of galactose intolerance, lactase deficiency, or glucose-galactose malabsorption;
3. Pregnancy;
4. In women: insufficiently reliable contraception methods (requirement: Pearl Index <1);
5. Lactation;
6. Known renal impairment (creatinine clearance <30 mL/min);
7. Known liver disease (Child-Pugh score B and C);
8. Known ulcerative gastrointestinal disorders within 30 days before treatment start or during treatment;
9. Uncontrolled, severe arterial hypertension (stage 3);
10. Artificial heart valves;
11. Acute pulmonary embolism;
12. Bronchiectasis or pulmonary bleeding in the patient medical history;
13. Known vascular retinopathy;
14. Intracranial or intracerebral hemorrhage within 30 days before trial start or during trial;
15. Brain, spinal cord, or eye surgery within the 30 days before trial start or during trial;
16. Spinal/epidural anesthesia or puncture within 2 weeks before treatment or during trial;
17. Administration of systemic heparin within 7 days before treatment;
18. Use of nonsteroid antirheumatic (NSAR) drugs or platelet aggregation inhibitors within 7 days before treatment or during trial;
19. Use of vitamin K antagonists (phenprocoumon, warfarin) and/or thrombin inhibitors (dabigatran) within 7 days before treatment or during trial;
20. Concomitant administration of CYP3A4 inductors (eg, rifampicin, phenytoin, carbamazepine, phenobarbital, and St. John's wort);
21. Concomitant systemic treatment with azole antifungals (eg, ketoconazole, itraconazole, voriconazole, and posaconazole);
22. Concomitant systemic treatment with human immunodeficiency virus (HIV) protease inhibitors (eg, ritonavir); and
23. Concomitant systemic treatment with dronedarone.

Selection of Study Centers

Study centers were selected according to their focus on the indication being studied and their clinical experience treating patients with livedoid vasculopathy. Because facilities

specialized in the treatment of this disease are rare in Germany, it is expected that patients from a wider radius will attend the study centers.

Treatment Schedule

Patients will take 1 film-coated tablet of rivaroxaban orally with food each morning and evening for 12 weeks. After 7 pain-free days, the dose will be reduced to 1 tablet each morning. If pain returns, the dose will immediately be increased to the original dose of 1 tablet each morning and evening.

Schedule of Investigations (Visits)

A total of 4 visits are foreseen: at baseline, 2 interim visits after 4 and 8 weeks, and a final visit after 12 weeks. Deviation of 2 days is allowed. A shortening or lengthening of the interval before a visit will be compensated to maintain the 4-week examination rhythm.

Entries in patient diaries will be made on a daily basis and, if possible, at the same time each day. All entries have to refer to the past 24 hours. Documentation will start on the day of first tablet intake (start of therapy) and will end after 12 weeks.

End of Interventional Patient Treatment

The regular interventional treatment for a single patient ends with the last visit after 12 weeks.

The patient is considered to have prematurely terminated the study if at least 1 of the following criteria is met:

1. Withdrawal of informed consent
2. Violation of inclusion or exclusion criteria during the study
3. If the investigator deems that further participation of the patient is not justifiable
4. Lack of compliance
5. Premature termination of the complete study
6. A decrease in hemoglobin level of 2 units from baseline, if bleeding cannot be stopped

Further treatment of patients with the study drug after the trial ends is not planned. The optimal treatment regimen for the patient will be discussed.

Continuous medical attendance is granted because only study centers with a focus on the treatment of livedoid vasculopathy will be selected.

Stopping Rules

The study as a whole will end when all queries generated by the corresponding data management department are resolved and the study database is closed, but not later than 4 months after the last patient's last visit.

In case of major contraventions of the AMG, data protection regulations, or of the principles laid down in the International Conference on Harmonisation of Technical Requirements for Registration of Pharmaceuticals for Human Use (ICH) good clinical practice guidelines (ICH E6), a study center may have to terminate the study prematurely.

Premature termination of the study as a whole will be taken into consideration if:

1. Individual protocol violations accumulate
2. Ethical or scientific justification is compromised or no longer valid
3. Serious adverse reactions occur in a large segment of the patient population, suggesting that patient safety according to the risk risk-benefit assessment can no longer be guaranteed
4. Protocol violations compromise the scientific integrity of the study with regard to planned statistical analysis or other aspects
5. Prerequisites for proper study conduct are no longer fulfilled for other reasons

Premature termination of the trial requires joint agreement between the principal investigator (sponsor) and the corresponding biostatistician. The decision has to be justified in writing.

The study may be stopped during consultations. All other study-related procedures (eg, examinations, study visits, documentation) may be continued. Therapy will be continued in a symptom-guided manner and documented according to protocol.

It has to be considered that premature termination of study treatment may lead to irrecoverable loss of patient data. The decision regarding premature termination of the study as a whole has to be jointly taken by the sponsor and the corresponding biostatistician.

Patient Safety Monitoring

Collection and documentation of adverse events (AEs) begins when the patient signs the informed consent form and ends on the date of the last study visit for each patient (individual end of study).

All AEs have to be documented in the patient file and on the AE form of the case report forms (CRF) immediately. The investigator has to report any serious adverse event to the sponsor immediately (no longer than 24 hours after awareness).

In case of a fatal or life-threatening suspected unexpected serious adverse reaction (SUSAR), the sponsor must report relevant information immediately or within 7 days of detection to the competent ethics committee, any competent authorities and the study centers involved according to applicable legal regulations. Additional relevant information has to be provided within an additional period of 8 days.

All other cases of SUSARs that come to the attention of the sponsor must be reported immediately or within 15 days to the corresponding ethics committee, authorities, and study centers involved according to applicable legal regulations.

Annual safety update reports are to be made according to applicable legal regulations on a yearly basis or as requested, and are to be provided to the competent authorities and corresponding ethics committee.

Documentation

Data will be recorded using CRFs. The investigator is responsible for the timely, correct, complete, and legible recording of study data in the CRFs. He/she will confirm correctness of the data by his/her signature.

CRFs are to be completed with black ballpoint pen. Corrections are to be documented as follows: the wrong entry will be crossed out with a single line and corrections will be entered next to the crossed-out text and verified by date and initials, stating the reason for the change if necessary. Instructions for use (entry and corrections) are included in each CRF.

Source data according to the ICH E6 guideline are original records of the patient file, doctor's letters, certified copies of original records, and laboratory printouts. In addition, all patient questionnaires (self-reporting) and patient diaries are regarded as source data.

Study data are to be recorded from the patient files. As an exception, self-reporting questionnaires and patient diaries for this study will be contained in the CRF only and not in the patient files. When specifically required, data from the patient diaries will have to be transferred to the CRF.

All patient data will anonymized. Each patient will be unambiguously identified by a patient identification number assigned at the study center. The investigator will keep a patient identification list documenting the patient identification number with the patient's full name, date of birth, sex, and date of informed consent.

The patient identification list is part of the investigator file and will remain at the study site. The patient identification number consists of a 1-digit center number, a hyphen, and a 2-digit patient number assigned for each center.

Assurance of Data Quality

The CRFs and questionnaires will be provided with carbon paper copies. The original sheets will either be collected by the study monitor during monitoring visits and forwarded to the study coordinator, or directly requested by the study coordinator. At the study coordination center, the CRFs will be checked for completeness and consistency (in-house review). Queries will be generated for missing or implausible entries and sent to the study centers. After clarification of implausible entries and completion of missing data, CRFs and questionnaires will be handed over to the corresponding data management department.

The data management department carries out data entry with the validated study software MACRO. Data entry will be performed by 2 individuals independently. Databases resulting from first and second entries will be compared. Additional plausibility checks will be conducted regarding ranges, validity, and consistency. In case of nonplausible data, the study coordinator will be informed and he/she will forward queries to the study centers and request clarification. Answers to queries will be filed together with CRFs.

At the end of the study and after correction of all implausible data, the database will be closed. Closure of the database will be documented. The study database will be handed over to the corresponding statistics department.

Quality Control and Quality Assurance

Quality control of the study is assured by monitoring in the study centers involved. For each monitoring visit, a monitoring report will be generated documenting the progress of the study and describing actual problems of study conduct. The explicit mode and extent of monitoring is described in a separate monitoring manual.

All investigators will declare their consent to regular visits of study monitors at the study centers. In addition, they must provide direct access to all relevant study documents including original patient documents relevant to the study.

The sponsor or designated auditors are entitled to conduct audits at the study center and other facilities participating in the study. They are entitled to inspect and review all study-relevant documents.

Ethical and Regulatory Aspects

The study will be conducted in compliance with the current version of the Declaration of Helsinki. The present study will not begin before approval has been obtained from the leading ethics committee. The study will not be started at any additional study sites before the corresponding local ethics committee has confirmed the adequacy of the study site and the investigators.

Before inclusion in the study, the investigator will inform each patient about the nature, significance, risks, and scope of the study, as well as patients' right to withdraw from the study at any time without prejudice. Patients will receive an informed consent form describing the study in nonscientific and generally understandable language.

The patient has to consent to study participation in writing. The investigator has to provide ample time for the patient to make a decision and allow for clarification of any questions before the consent form is signed.

According to the German drug law AMG § 40 (2a), the patients will be informed that their pseudonymized disease-related data will be stored in the study database and evaluated for scientific purposes. They have to consent to the use of their pseudonymized data in writing.

The informed consent form is to be signed by the patient and by the investigator of the study site. The patient information and consent form will be kept as 2 official (signed) copies. One copy will remain with the investigator; the other copy will be given to the patient.

This clinical study is to be carried out in conformity with the requirements of the current German drug law (AMG), all applicable legal provisions regarding data protection, and principles of good clinical practice.

The clinical study will not begin before its approval by the competent federal authority. In case of substantial amendments, a new application will be submitted. The amendment can be implemented only after approval by the competent federal authority. According to §67 AMG, the investigator must notify the local supervisory authority regarding the beginning of the study, its regular or premature termination, and any amendments. These notifications are to be made by the sponsor or an appointed designee of the sponsor on behalf of the investigator.

For the current clinical study, an insurance policy is provided and a copy of the general insurance terms and conditions will be given to the patients.

Original central study documents including CRFs have to be archived by the sponsor for at least 10 years after the end of the study. The study investigator must archive all documents of the investigator's file and copies of the CRFs for the previously mentioned period.

After biometrical analysis, the principal investigator will generate a clinical study report (CSR). The CSR includes the clinical report, the statistical report, individual patient data listings, and conclusions. The report is to be signed by the principal investigator and by the biostatistician. Publication of the study results is the responsibility of the principal investigator irrespective of study results. A summary of the CSR has to be provided to the competent ethics committee and to the competent authority within 12 months after the end of the study. Compliance with this study protocol is mandatory. Each deviation from time schedule, scheduled study procedures, or study treatments initiated by the investigator has to be documented and justified (eg, emergency measures).

Results

Enrollment started in December 2012 and was still open at the date of submission. The study is expected to finish in November 2014.

Discussion

Rationale for This Trial

Livedoid vasculopathy is associated with increased thrombophilia in the cutaneous microcirculation and the continuous use of anticoagulants helps improve the symptoms [4]. The causes of cutaneous infarctions are varied, but ultimately follow the known mechanisms of the coagulation cascade. Low-molecular-weight heparins have been shown to be effective in the treatment of livedoid vasculopathy [1,13,23].

In 2009, a new drug became available for the preventive treatment of thrombosis: the direct factor Xa inhibitor, rivaroxaban (Xarelto) [34]. Depending on the dose, rivaroxaban can have different applications. At a dose of 10 mg, prevention of venous thromboembolism in adult patients who have undergone elective hip or knee joint replacement surgery. At a dose of 15-20 mg, prevention of strokes and systemic embolism in adult patients with nonvalvular atrial fibrillation and 1 or more risk factors, such as age older than 75 years or a history of congestive heart failure, hypertension, diabetes, stroke, or transient ischemic attacks; and treatment of deep vein thrombosis (DVT) and prevention of chronic DVT and pulmonary embolism after acute DVT in adults.

Rivaroxaban affects the coagulation cascade and inhibits the factor Xa–dependent conversion of prothrombin to thrombin, thereby considerably reducing the risk of thrombosis. This mechanism has been applied successfully in isolated cases to treat livedoid vasculopathy in the skin. Therefore, it is expected that this oral treatment will help prevent cutaneous infarctions in livedoid vasculopathy patients and lead to an increase in their quality of life.

This leads to the question: How effective is rivaroxaban in treating livedoid vasculopathy?

Risk-Benefit Analysis

The essentially prothrombotic nature of livedoid vasculopathy has been sufficiently documented and supports the use of anticoagulants in a therapeutic context [1,13,23]. The nonbinding, consensus-based application of heparin injections used until now improves the symptoms of livedoid vasculopathy, but constitutes a significant burden for patients due to the constant administration of subcutaneous injections. In addition, the effectiveness of heparin has not been demonstrated in clinical trials. The development of new oral anticoagulants is, therefore, a relevant contribution to the treatment of the disease. Treatment of individual cases has already shown the effectiveness of this therapy and participants will receive a known IMP that has already been tested on more than 16,000 patients [34]. Associated risks primarily involve a higher chance and severity of hemorrhage. However, these risks are related to anticoagulants in general and are not drug-specific. Updated indications show that—in view of safety considerations and the associated risk-benefit analysis—rivaroxaban is considered appropriate for treating severely ill patients. This includes the prevention of strokes and systemic embolism in adult patients with nonvalvular atrial fibrillation and 1 or more risk factors, such as age older than 75 years or a history of congestive heart failure, hypertension, diabetes, stroke, or transient ischemic attacks.

To increase patient safety, the trial will exclude individuals with diseases and ailments associated with a negative reaction to rivaroxaban (eg, kidney disorders, patients with increased risk of hemorrhage). In addition, administration of any concomitant medications that could produce an uncontrolled increase or decrease of rivaroxaban concentrations in plasma will not be allowed to (1) reduce the risk of hemorrhage and (2) not exceed the trial dose and mask a possible therapeutic effect. Approved treatment alternatives do not exist.

Main drawbacks for participants in the trial include the increased amount of time they will have to invest in keeping a patient diary and providing additional information (eg, surveys, questionnaires) to the research team. However, in view of the expected improvement in patient quality of life, both of these drawbacks seem acceptable.

Discussion of the Inclusion and Exclusion Criteria

The innocuousness and effectiveness of rivaroxaban has not been proven in children aged 0-18 years and no studies for this age group exist. Therefore, only patients older than 18 years will be included in the trial.

The maximum age limit of 80 years was chosen because rivaroxaban can also be indicated for patients older than age 75 years. However, the research team must take into account that older patients, compared to younger patients, show higher plasma concentrations (mean area under the curve increase of 1.5 fold) in tests. This is primarily caused by a (apparent) reduction in total body and renal clearance. An adjustment of the dose is not considered necessary.

Because there are no data regarding the use of rivaroxaban in pregnant women, it will be necessary to ensure that reliable contraceptive measures are used. There is also a lack of data on the use of the drug during lactation. In addition, experimental animal models have indicated the transfer of rivaroxaban into breast milk. Therefore, rivaroxaban is contraindicated during breastfeeding.

The use of drugs that cause a clinically significant increase or reduction in plasma concentrations of rivaroxaban is considered an exclusion criterion for this trial.

Diseases and complaints known to have potential adverse effects on patients receiving rivaroxaban (eg, kidney malfunction, risk of hemorrhage) are also exclusion criteria for the present trial.

Rationale for the Dosage and Treatment Duration

Depending on the dose, rivaroxaban is approved mainly for prophylactic use. The recommended dose is 15-20 mg per day. In the case of this trial, a treatment dose of 20 mg was chosen to promote more rapid pain relief in patients. This course of action is also common in the off-label use of low-molecular-weight heparins (eg, enoxaparin, 1-2 mg/kg body weight) and is widely followed in Europe because of lack of approved treatment alternatives.

Rivaroxaban has a terminal half-life of 5-13 hours and peak plasma concentration is reached after 2-4 hours [35]. To ensure the highest possible degree of therapeutic uniformity over 24 hours, the daily dose is divided into 2 doses (10 mg in the morning and 10 mg in the evening). This also seeks to prevent unnecessary peak concentrations and a higher associated risk of hemorrhage.

At 7 days after the onset of pain relief, drug intake is reduced to a maintenance dose of 10 mg per day. Considering the relationship between dose and adverse reactions, the aim is to achieve therapeutic effectiveness with the lowest possible prophylactic dose, thus further reducing the risk of adverse effects.

As described in the Introduction, livedoid vasculopathy is an incurable genetic disorder that recurs if treatment is stopped. This explains the relatively long treatment duration of 12 weeks to study the sustained therapeutic effect of the drug and document the complete regeneration and epithelialization of skin wounds over the course of the trial.

Limitations

In the present study, pain is assessed for testing the efficacy of rivaroxaban. It would be desirable to directly measure the increased microcirculatory perfusion as an objective marker for treatment efficacy.

The calculation of the study sample size is based on assumptions found in case reports of this rare disease. The sample size was calculated for a power of 80%.

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Authors' Contributions

AD and TG designed the study protocol and AD, CH, and TG prepared the manuscript.

Conflicts of Interest

None declared.

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Abbreviations

AE: adverse event
AMG: Arzneimittelgesetz (German drug law)
AR: adverse reaction
CRF: case report form
CSR: clinical study report
DLQI: Dermatology Life Quality Index
DVT: deep vein thrombosis
ICH: International Conference on Harmonisation of Technical Requirements for Registration of Pharmaceuticals for Human Use
IMP: investigational medicinal product
ITT: intention-to-treat
PP: per-protocol
SUSAR: suspected unexpected serious adverse reaction
VAS: visual analog scale

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Original Paper

Development and Validation of Questionnaires Exploring Health Care Professionals' Intention to Use Wiki-Based Reminders to Promote Best Practices in Trauma

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Abstract

Background: Little is known about factors influencing professionals' use of wikis.

Objective: We developed and validated two questionnaires to assess health care professionals' intention to use wiki-based reminders for the management of trauma patients.

Methods: We developed questionnaires for emergency physicians (EPs) and allied health professions (AHPs) based on the Theory of Planned Behavior and adapted them to the salient beliefs of each, identified in an earlier study. Items measured demographics and direct and indirect theoretical constructs. We piloted the questionnaires with 2 focus groups (5 EPs and 5 AHPs) to identify problems of wording and length. Based on feedback, we adjusted the wording and combined certain items. A new convenience sample of 25 EPs and 26 AHPs then performed a test-retest of the questionnaires at a 2-week interval. We assessed internal consistency using Cronbach alpha coefficients and temporal stability of items with an agreement intraclass correlation coefficient (ICC).

Results: Five EPs and 5 AHPs (3 nurses, 1 respiratory therapist, and 1 pharmacist) formed 2 focus groups; 25 EPs and 26 AHPs (12 nurses, 7 respiratory therapists, and 7 pharmacists) completed the test and retest. The EP questionnaire test-retest scores for consistency (Cronbach alpha) and stability (ICC) were intention (test: Cronbach alpha=.94; retest: Cronbach alpha=.98; ICC=.89), attitude (.74, .72, .70), subjective norm (.79, .78, .75), perceived behavioral control (.67, .65, .66), attitudinal beliefs (.94, .86, .60), normative beliefs (.83, .87, .79), and control beliefs barriers (.58, .67, .78) and facilitators (.97, .85, .30). The AHP questionnaire scores for consistency and stability were: intention (test Cronbach alpha=.69, retest Cronbach alpha=.81, ICC=.48), attitude (.85, .87, .83), subjective norm (.47, .82, .62), perceived behavioral control (.55, .62, .60), attitudinal beliefs (.92, .91, .82), normative beliefs (.85, .90, .74), and control beliefs barriers (.58, .55, .66) and facilitators (.72, .94, –.05). To improve the psychometric properties of both questionnaires, we reformulated poorly consistent or unstable items.

Conclusions: Our new theory-based questionnaires to measure health care professionals' intention to use wiki-based reminders have adequate validity and reliability for use in large surveys. In the long run, they can be used to develop a theory-based implementation intervention for a wiki promoting best practices in trauma care.

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KEYWORDS

knowledge translation; wiki; collaborative writing applications; decision support tools; health informatics; Theory of Planned Behavior; trauma care; traumatic brain injury; interprofessional collaboration

Introduction

Clinical practice does not always reflect best evidence. High proportions of inappropriate care have been reported in different health care systems and settings [1]. This has a huge impact on both patient outcomes and health care costs. As passive dissemination of evidence has not proven adequate for encouraging implementation of research-based recommendations for changes in practice, new strategies are being advocated [2].

Information and communication technologies (ICTs) such as computerized decision support systems have been suggested as a possible solution for improving research uptake and increasing evidence-based practice [3]. Aiming to improve care and reduce costs, governments have invested billions of dollars to implement ICTs, including decision support systems, but these systems have yet to deliver the expected benefits [4]. Moreover, some health care professionals have rejected ICTs on the grounds that they are slow, incompatible with work processes, difficult to access, costly to implement, and cannot be adapted to local practices [4-10]. Furthermore, local initiatives to adapt various ICT solutions seem to be restricted to a small number of hospitals and tools are mostly designed for local use only [11-13]. Transfer of these local initiatives to the larger health care community is often slow and complex. In emergency departments (EDs), where shift work is prevalent, getting health care professionals to collaborate in creating, using, and updating decision support tools (eg, care protocols, care pathways, and decision aids) is particularly difficult [14]. These decision support tools can be translated into paper-based or computer-based reminders that support clinicians' or patients' decision making at the bedside. The most important factors influencing the creation, use, and updating of any form of reminders to promote best practices may be time and collaboration within and across care teams [15,16]. Wikis are an open-source and low-cost means of accelerating innovation and permitting a broad spectrum of stakeholders to collaborate efficiently for this purpose.

Wikis are knowledge management platforms that empower stakeholders to implement evidence-based decision support tools in different areas of health care [17]. A wiki is a website that uses a novel technology to allow people to view and edit website content, with viewing and editing privileges determined by various levels of access. Wikipedia—the best-known wiki—has 365 million visitors per month, is the sixth most popular website in the world, and its medical articles (available in 271 languages) are viewed approximately 150 million times per month [18]. Many health organizations have started using wikis to manage knowledge and coordinate care [19-23]. A recent scoping review found that wikis are effective educational interventions for health students and professionals and that they have many positive impacts on knowledge translation processes and outcomes: theoretical behavioral change domains (eg, beliefs about capabilities), learning (eg, skills and knowledge), communication, collaboration, knowledge management, health care efficiency, quality improvement, and disease prevention [17].

A wiki could permit stakeholders in 1 or many EDs to collaborate asynchronously in the updating and creation of reminders, decreasing duplication efforts and reducing the time needed. However, despite increasing evidence supporting the use of wikis in various settings, there is a lack of knowledge about the factors influencing professionals' use of wikis and about how best to implement them in health care settings [24,25].

The objectives of this study were to develop and test the psychometric properties of 2 questionnaires based on the Theory of Planned Behavior (TPB) [26] exploring the intention of ED health care professionals and the determinants of this intention to use wiki-based reminders promoting best practices for the management of severe traumatic brain injury (TBI) victims.

Methods

Study Design

The protocol for this mixed methods study describes 4 phases [27]: (1) eliciting salient beliefs [25], (2) developing the questionnaires, (3) piloting the questionnaires, and (4) test-retest of the adjusted questionnaires. Phase 1 of this project identified ED professionals' salient beliefs concerning the use of wiki-based reminders promoting best practices for the management of severe TBI [25]. The current study represents the later phases (2, 3, and 4) of the published research protocol [27]. Our participants, emergency physicians (EPs) and allied health professionals (AHPs), came from 3 hospitals of 3 different trauma levels (I, II, and III) in the province of Quebec, Canada. All our participants were French speaking. The ethics committees from the 3 hospitals approved this study and there were no financial incentives offered to participants.

Definition of the Behavior for the Present Survey

We chose to study the intention (and determinants of intention) of ED health care professionals to use a wiki-based reminder promoting best practices for the management of severe TBI victims in the ED in the Province of Quebec, Canada. Definition of the behavior was:

1. Action: to use

2. Target: a wiki-based reminder promoting best practice
3. Context: management of severe TBI victims in EDs in the province of Quebec, Canada

Phase 1: Elicitation of Salient Beliefs

A complete report of Phase 1 of our study has been published [25]. In summary, we conducted semistructured interviews to elicit EPs' and AHPs' beliefs about using a wiki-based reminder. In order to clearly depict the behavior being studied, 4 videos were created presenting 4 different health care professionals (emergency physician, nurse, respiratory therapist, and pharmacist) performing the behavior (see Multimedia Appendix 1 to access the YouTube videos in French). After watching the video specific to their profession, each participant was interviewed about their behavioral, control, and normative beliefs (ie, what they saw as advantages, disadvantages, barriers, and facilitators to their use of a wiki-based reminder) and how they felt important referents would perceive their use of a wiki-based reminder. After ranking each belief from the most reported to the least reported, we considered the top 75% most-reported beliefs as salient. We also retained certain beliefs as salient although they were not among the top 75% most reported. This decision was based on our knowledge of the literature, our experience in implementing care protocols for trauma, or our fear of excluding important negative beliefs. This study generated 2 different sets of salient beliefs for EPs and AHPs that were used to construct 2 different questionnaires.

Phase 2: Questionnaire Development

Direct Construct Items

In both questionnaires, we included items to measure the constructs identified in our theoretical model: intention (n=3), perceived behavioral control (n=3), attitude (n=4), and subjective norm (n=3). The items were formulated so that participants could then evaluate their level of agreement with each statement on a 7-point Likert scale.

Indirect Construct Items

We selected the salient behavioral, normative, and control beliefs identified in our published Phase I and converted these into a set of statements for each questionnaire. The items were formulated so that participants could evaluate their level of agreement on a 7-point Likert scale about each advantage, disadvantage, positive referent, negative referent, barrier, and facilitator presented.

Characteristics of Health Care Professionals

We assessed the following demographic characteristics: age, gender, type of health care professional, and diploma (AHP questionnaire), training level of EPs (EP questionnaire), type of health care center (level I, II, III), number of years in practice, presence of computers with unrestricted access to the Internet within their ED, availability of Wi-Fi for professionals, availability of Wi-Fi for patients, previous consultation of or contribution to a wiki, and membership in local trauma committees.

Ordering of Questions in the Questionnaires

The drafts of our initial questionnaires were created without randomly mixing the items and in the following order: intention, perceived behavioral control, subjective norm, attitude, control beliefs (facilitators), control beliefs (barriers), normative beliefs, and attitudinal beliefs (advantages and disadvantages).

Phase 3: Pilot Testing of the Questionnaires

We pilot tested our questionnaires by asking a convenience sample of 10 participants (5 physicians and 5 AHPs) from our population to answer the questionnaire intended for their own professional group before the focus group. Participants for this phase were recruited purposefully to represent a wide range of professionals and to represent different age groups. Participants could choose either a paper-based survey or a Web-based survey (SurveyMonkey). This choice was meant to ensure that participants less comfortable with computers could access a paper-based version of our questionnaire. We formed 2 focus groups (EPs in 1 and AHPs in the other) to tell us whether they had any difficulty answering the questions. The Web-based survey contained an HTML link to a YouTube video presenting the behavior being studied and paper-based participants were sent the link in the invitation email. For participants who had not completed the survey before the focus group, we presented the video during the focus groups and then they answered a paper-based questionnaire.

We then interviewed participants about both the Web-based and paper-based questionnaires to check comprehension and clarity. Interviews were based on a cognitive interview methodology using a preplanned questionnaire (see Multimedia Appendix 2) [28]. Focus group participants were asked to (1) read the instructions and tell us what they understood, (2) specify what our questions meant to them, (3) specify what the studied behavior meant to them and what a wiki-based reminder represented to them, (4) identify any ambiguous or complex terms, (5) evaluate their ease or difficulty in answering our questions and examine the difficulties, (6) identify the questions that were the most difficult to understand, (7) specify if each answer option was clearly different from the others and, if not, identify those that were too similar, and (8) suggest changing answer options that were ambiguous or that did not translate their thought processes adequately. We also asked questions about the questionnaire length and consequent participant fatigue. Focus groups were recorded, transcribed verbatim, and then analyzed to identify the adjustments that were needed to the wording of some items and to the visual presentation of the questionnaires.

Phase 4: Test and Retest

Once adjustments based on the focus groups' comments were made, a test-retest study of the revised questionnaires was done with a different convenience sample of 25 EPs and 26 AHPs. These participants had not participated in the elicitation phase (Phase 1) or in the focus groups (Phase 2), but worked in the same 3 trauma centers (levels I, II, and III). The EP participants were recruited from these 3 EDs after presenting this project at a monthly departmental meeting in each center. The AHP participants were recruited after contacting the head of each

service and asking them to identify potential participants. Consent to participate in this test-retest study was obtained from all participants after explaining the length of the questionnaire and they were informed that all personal information would remain confidential. However, participants were assigned a unique identifier code to write on their questionnaires to link their test and retest responses. This unique identifier was stored separately from the results. The same questionnaire was performed 2 weeks later with the same participants (retest). For this phase, we again allowed participants to choose between the paper-based and the Web-based questionnaire, but they had to use the same modality for both the test and retest. SurveyMonkey automatically collected the data for the Web version, but responses were manually entered in a spreadsheet for the paper-based questionnaires. The Web-based questionnaire did not contain an official completeness check to identify unanswered items; however, participants were allowed to review all their responses by returning to previous items and participants were asked at the last page to complete all items. We monitored duplicate participation with the unique identifier provided.

Data Analysis

Simple descriptive statistics were used to compare demographic information for EP and AHP participants in our test-retest sample. We also used simple descriptive statistics to compare the demographics of the participants who used the paper-based vs the Web-based questionnaire. We conducted *t* tests for normally distributed continuous variables and Wilcoxon-Mann-Whitney tests for continuous variables that were not normally distributed. For all categorical variables, we used the Fisher exact test. The internal consistency of the constructs (the tendency of answers within a group of constructs) was measured using Cronbach alpha coefficient. To measure stability over time in the constructs, an agreement intraclass correlation coefficient (ICC) was measured. We used criteria published by Landis and Koch to determine the level of consistency and reproducibility of our items [29]. For any missing data for single questionnaire items, we imputed the average of the other items measuring the same construct. Statistical analysis was performed by a biostatistician using SAS version 9.3 (SAS Institute Inc., Cary, NC, USA).

Results

Participants

The number of participants per phase of the study is presented in a flow diagram (Figure 1).

Demographic characteristics of the focus groups and test-retest participants are presented in Table 1.

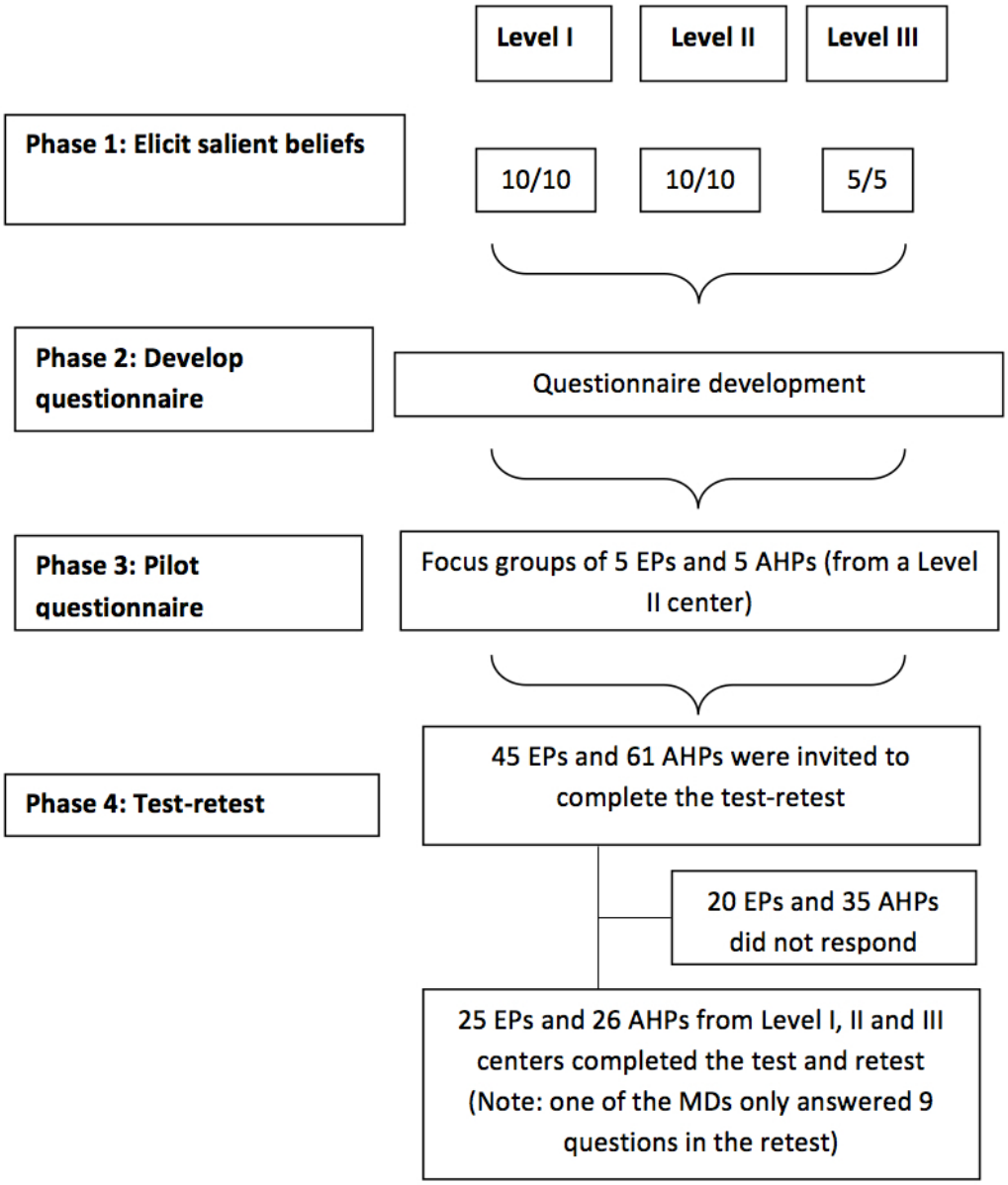
Five EPs and 5 AHPs (3 nurses, 1 respiratory therapist, and 1 pharmacist) formed the focus groups. Among all the test-retest participants, 25% (13/51) came from the level I trauma center, 63% (32/51) from the level II and 12% (6/51) from the level III. Among the 26 test-retest AHPs, there were 12 nurses, 7 respiratory therapists, and 7 pharmacists. Most participating EPs were certified in Emergency Medicine by the College of Family Physicians of Canada (84%, 21/25), 12% (3/25) were certified by the Royal College of Physicians of Canada, and 1 was a family physician certified by the College of Family Physicians of Canada without any Emergency Medicine certification. Compared to the AHPs, the EPs participating in the test-retest were more likely to be older (P=.03), male (P<.001), to have better access to the Internet (P=.02) and to report a higher prevalence of wiki use for personal purposes (72% vs 31%, P=.005). Although more EPs tended to use wikis for professional purposes (20%) than AHPs (8%), this difference was not significant. None of our test-retest participants had ever edited a wiki before. However, in our focus groups 1 EP and 1 AHP had edited a wiki.

Table 1. Demographic characteristics of the focus groups and test-retest participants.

Variables	Focus groups		Test-retest participants		
	EPs (n=5)	AHPs (n=5)	EPs (n=25)	AHPs (n=26)	*P* value[a]
Age (years)					
Mean (SD)	49 (10)	36 (3)	41.5 (8.9)	35.5 (11.6)	.03
Median (IQR)	48 (41-56)	36 (35-38)	42 (34-48)	33 (26-43)	
Gender, n (%)					
Male	4 (80)	2 (40)	19 (76)	3 (12)	<.001
Female	1 (20)	3 (60)	6 (24)	23 (88)	
Trauma center level, n (%)					.23
I	0	0	6 (24)	7 (27)	
II	5 (100)	5 (100)	18 (72)	14 (54)	
III	0	0	1 (4)	5 (19)	
Profession, n (%)					
Physician	5 (100)		25 (100)		
Nurse		3 (60)		12 (46)	
Respiratory therapist		1 (20)		7 (27)	
Pharmacist		1 (20)		7 (27)	
Emergency medicine certification, n (%)		N/A		N/A	
College of Family Physicians	2 (40)		21 (84)		
Royal College of Physicians of Canada	3 (60)		3 (12)		
Other	0		1 (4)		
Clinical experience (years)					
Mean (SD)	15 (9)	12 (4)	12.5 (9.4)	13.2 (11.1)	.79
Median (IQR)	13 (8-18)	12 (11-14)	11 (5.5-17)	10.8 (5-19.8)	
Internet access in the ED, n (%)	5 (100)	3 (60)	25 (100%)	20 (77)	.02
Professional use of a wiki, n (%)	2 (40)	2 (40)	5 (20)	2 (8)	.25
Personal use of a wiki, n (%)	4 (80)	2 (40)	18 (72)	8 (31)	.005
Previous editing of a wiki, n (%)	1 (20)	1 (20)	0 (0)	0 (0)	>.99

[a]*P* values were only calculated for the test-retest group.

Figure 1. Number of participants per phase of the study. EPs: physicians; AHPs: allied health professionals.



Phase 2: Number and Content of Items in the First Version of the Questionnaires

In the EP questionnaire, there were a total of 13 pages with 63 items: 13 direct construct items, 35 indirect construct items, and 15 demographic characteristic items. For the AHP questionnaire, we created 11 pages with 58 items: 13 direct construct items, 31 indirect construct items, and 14 demographic characteristic items. The original versions of the questionnaires developed during Phase 2 are available in French (see Multimedia Appendices 3 and 4).

Phase 3: Focus Group Comments and Changes Made to the Questionnaires

In the focus groups, all EPs chose to answer the Web-based survey rather than the paper-based version. The reason evoked was that it was easier to access the survey after receiving the email invitation. One AHP chose the paper version because this participant had not filled out the questionnaire before the focus group and all the other AHPs used the Web-based questionnaire before attending the focus group. Multimedia Appendix 5 lists all the comments made by our focus group participants and the changes we made to the questionnaire in consequence. In summary, we changed the wording of certain items, reduced the length of the questionnaire (without reducing any of the TPB items), removed 1 item from our demographic questions (a question about Wi-Fi availability), and clarified certain items to make the questionnaire easier to complete.

Phase 4: Test-Retest Results and Changes Made to the Questionnaires

Table 2 compares the characteristics of participants who chose the Web-based questionnaire with those of participants who chose the paper-based questionnaire.

Most EPs (72%, 18/25) used the paper-based version. In contrast, most AHPs (92%, 24/26) preferred the Web-based version for the test-retest and only 2 of 26 AHPs (8%) chose to

use the paper-based version. There were no significant differences in characteristics between EPs who used the Web-based vs the paper-based questionnaire. However, 16 of 18 EPs who used the paper-based version did so because the survey was presented during a monthly department meeting when Internet access was not available. Among EPs who had the choice between paper and Web, 6 of 9 (67%) opted for the Web-based questionnaire. The 2 AHPs who chose the paper version worked at the level III trauma center and did not have Internet access in their hospital. All participants used the same administration mode (paper vs Web) for the test and retest. All participants completed the test and retest except 1 EP who only completed 9 items in the retest questionnaire and did not complete it. The data for these 9 items (all relating to direct TPB questionnaire items) were retained in our analysis, but no imputation was performed for the missing data for this participant. There were no duplicate participants.

Table 2. Demographic characteristics of participants using Web-based questionnaire vs paper-based questionnaire.

Variables	Emergency physicians (n=25)[a]			Allied health professionals (n=26)		
	Paper (n=18)	Web (n=7)	*P*	Paper (n=2)	Web (n=24)	*P*
Age (years)						
Mean (SD)	41.6 (8.6)	41.3 (10.5)	.95	43.5 (20.5)	34.8 (11.4)	.44
Median (IQR)	42 (36-47)	41 (32-49)		44 (32-49)	33 (24-35)	
Gender, n (%)			.74			.60
Male	14 (78)	5 (71)		0 (0)	3 (12.5)	
Female	4 (22)	2 (29)		2 (100)	21 (87.5)	
Trauma center level, n (%)			.007			.003
I	2 (11)	4 (57)		0 (0)	7 (29)	
II	16 (88)	2 (29)		0 (0)	14 (58)	
III	0 (0)	1 (14)		2 (100)	3 (12.5)	
Certification, n (%)			.11		N/A	
College of Family Physicians	16 (88)	4 (57)				
Royal College of Physicians	2 (11)	2 (29)				
Other	0 (0)	1 (14)				
Profession, n (%)						.13
Physician	18 (100)	7 (100)				
Nurse				0 (0)	12 (50)	
Respiratory therapist				2 (100)	5 (20)	
Pharmacist				0 (0)	7 (29)	
Clinical experience (years)						
Mean (SD)	12.2 (9.1)	13.1 (11.1)	.88	21 (22.6)	12.6 (10.2)	.55
Median (IQR)	11 (5-18)	11 (5-20)		21 (13-29)	11 (5-17)	
Internet access in ED, n (%)	18 (100)	7 (100)	>.99	0 (0)	20 (83%)	.046
Professional use of a wiki, n (%)	4 (22)	1 (14)	.66	1 (50)	1 (4)	.15
Personal use of a wiki, n (%)	12 (66)	6 (86)	.63	2 (100)	6 (25)	.09
Previous editing of a wiki, n (%)	0 (0)	0 (0)	>.99	0 (0)	0 (0)	>.99

[a]16 EPs were asked to use the paper-based survey and the 9 others could choose Web- or paper-based. All AHPs had the choice to use either Web- or paper-based.

Overall psychometric properties of our 2 questionnaires are presented in Table 3. Internal consistency and temporal stability for each individual item in both questionnaires are presented in Multimedia Appendix 6.

The internal consistency of the items in the EP questionnaire was high (Cronbach alpha >.8) for 4 constructs (intention, attitudinal beliefs, normative beliefs, and control belief facilitators), substantial (Cronbach alpha=.6-.8) for 3 constructs (attitude, perceived behavioral control, and subjective norm),

and moderate (Cronbach alpha=.4-.6) for control belief barriers (test Cronbach alpha=58; retest Cronbach alpha=.67). One item measuring the attitude construct was removed due to lack of consistency (test: Cronbach alpha=.06; retest: Cronbach alpha=.02) (see Multimedia Appendix 6). This reduced the length of the questionnaire without affecting the internal consistency of this construct (3 items to measure attitude: test: .89; retest: .91). One item measuring perceived behavioral control was formulated differently because it had low consistency (test: Cronbach alpha=.20; retest: Cronbach alpha=.26) (see Multimedia Appendix 6). For the control beliefs, 2 items were reformulated to increase their internal consistency. All items were considered moderately stable over time (ICC >.4) except for the items measuring facilitators (ICC=.30). Consequently, we reformulated the 3 items concerning facilitators and removed items that represented beliefs that were less salient (based on their ranking in our Phase 1 study) to decrease the questionnaire length.

For the AHP questionnaire, internal consistency was high (Cronbach alpha >.8) for 3 constructs (attitude, attitudinal beliefs, and normative beliefs), substantial (Cronbach alpha=.6-.8) for 2 constructs (intention and control beliefs facilitators), and moderate for 3 constructs (subjective norm, perceived behavioral control, and control beliefs barriers). Therefore, we reformulated items with poor internal consistency: 1 item measuring intention, 1 item for perceived behavioral control, and 1 item for subjective norm. The stability of the items in our AHP questionnaire was good for most constructs except control beliefs facilitators (ICC= –.05). Because the consistency and stability of the items measuring control beliefs (barriers and facilitators) were low, we removed 2 items measuring control beliefs (1 barrier and 1 facilitator) that were not consistent or stable.

The changes made to all the items in our questionnaires are listed in Multimedia Appendices 7 (EPs) and 8 (AHPs) and the final versions (in French) are found in Multimedia Appendices 9 (EPs) and 10 (AHPs). English versions of the questionnaires are also available (Multimedia Appendices 11 and 12). The final EP questionnaire has 11 pages with 55 items and the AHP questionnaire has 9 pages containing 53 items.

Table 3. Overall internal consistency (Cronbach alpha) and temporal stability (intraclass correlation coefficient, ICC) of our questionnaire

Questionnaire constructs	Emergency physicians			Allied health professionals		
	Internal consistency		Stability	Internal consistency		Stability
	Test	Retest	ICC	Test	Retest	ICC
Intention	.94	.98	.89	.69	.81	.48[a]
Attitude	.74	.72[b]	.70	.85	.87	.83
Subjective norm	.79	.78	.75	.47[a]	.82	.62
Perceived behavioral control	.65	.67[a]	.68	.55[a]	.62	.60
Attitudinal beliefs	.94	.86	.60	.92	.91	.82
Normative beliefs	.83	.87	.80	.85	.90	.74
Control beliefs-barriers	.58	.67[c]	.78	.58[d]	.55	.66
Control beliefs-facilitators	.97	.85	.30[e]	.72	.94	–.05[f]

[a]1 item was reformulated.

[b]1 item was removed (with 3 items: test Cronbach alpha=.89, retest Cronbach alpha=.91, ICC=.78).

[c]2 items were reformulated.

[d]2 items were removed.

[e]3 items were reformulated and 2 removed.

[f]1 item was removed.

Discussion

Principal Results

The objectives of this study were to develop and test the psychometric properties of 2 questionnaires exploring the intention of ED health care professionals and the determinants of this intention to use wiki-based reminders promoting best practices for the management of severe TBI victims. Building on a previous study that had identified the salient beliefs of health care professionals about using wiki-based reminders, our 2 questionnaires will also measure the importance of each of these beliefs. The 4 videos developed in support of these 2 questionnaires helped the participants understand the behavior being investigated. The EP questionnaire now contains 55 items and the AHP questionnaire contains 53 items including the demographic items, as opposed to their original 68 and 58 items, respectively. Both questionnaires take approximately 10 minutes to complete after viewing a 6-minute video. Although some items needed reformulating, our questionnaires now have adequate validity and reliability for large surveys. These results lead us to the following observations.

First, to our knowledge, these questionnaires are among the first to be developed to understand how to implement a wiki that will promote best clinical practices in any area of health care.

Other authors have used the Technology Assessment Model to explore how health care professionals use and contribute to social media in general to share medical knowledge with other physicians in the medical community [24]. In contrast, we developed and validated our questionnaires by rigorously following the TPB methodology [30] and included all its constructs, both direct and indirect. As a result, our questionnaires will allow researchers to identify which behavioral determinants are most influential and, therefore, which determinants should be targeted when developing a theory-based intervention.

Second, these 2 questionnaires were developed and validated for 4 types of professionals (EPs, registered nurses, respiratory therapists, and pharmacists) and, thus, are ready for use across this range of health care professionals. The questionnaires are similar in terms of number of items, length of administration, and direct construct items, and many of the items investigating indirect construct items are similar (eg, validity of the information, hospital administration support, ease of access to the wiki-based reminder). However, other items exploring indirect constructs differ from 1 questionnaire to the other depending on the different salient beliefs held by each group of professionals (information captured in Phase 1). For example, the EP questionnaire contains an item investigating the influence of surgeons on their intention to use a wiki-based reminder to care for TBI victims, whereas the AHP questionnaire contains instead an item exploring the influence of quality control managers on their intention. Our results confirm our decision to begin our study by exploring salient beliefs and adapt each questionnaire accordingly. For example, our finding of higher current wiki use and Internet access among EPs than among AHPs supports the need for interventions adapted to each profession. Future investigations using our questionnaires will help us verify the importance of these factors among the different ED health care professionals and then construct profession-specific interventions to guide the implementation of a wiki promoting best practices in TBI trauma care.

Third, some of the constructs in our questionnaires lacked high levels of consistency (eg, perceived behavioral control and control beliefs), more so in the AHP questionnaire than in the EP questionnaire. One explanation for this lack of internal consistency is that the AHP participants were a more heterogeneous group than the EP participants. The AHPs were nurses, respiratory therapists, and pharmacists who all have different clinical tasks and who potentially perceive different levels of control over their clinical practice and behaviors. Moreover, although we tried to make the studied behavior as clear as possible by using profession-specific videos and repeating the description of the behavior in each question, it is still possible that participants did not all have the same behavior in mind. Future investigations with larger samples will help us verify these discrepancies between groups of professionals, and will be important to consider in a future implementation strategy.

Fourth, if given the choice, EPs and AHPs preferred to use the Web-based version rather than the paper version. Although our small sample size prevented us from comparing the internal consistency and stability of the Web-based version compared to the paper-based version, we must continue to have a paper version available because some professionals do not have Internet access (eg, AHPs and those working in smaller trauma centers). Offering a paper version will also allow us to capture the opinions of professionals who are not computer- or Web-savvy and yet are important stakeholders to consider in a future theory-based intervention. Most importantly, lack of Internet access among participants in this survey is an important barrier that must be addressed in any future wiki intervention. For our survey, we addressed this barrier by installing the survey videos on local hospital computers.

Fifth, the videos we created proved to be a useful tool for helping assess the intention of health care professionals to use wiki-based reminders. Using a video was advantageous in that the behavior being studied is new and complex (ie, to use a wiki-based reminder promoting best practice for the management of severe TBI victims in the ED in the Province of Quebec, Canada) and depends on many smaller microbehaviors (eg, logging on to the Internet, using a keyboard to type in the search terms to find the wiki-based reminder, checking off the appropriate prescriptions suggested by the wiki-based reminder). These videos allowed respondents to understand all the small implicit lead-in behaviors necessary to performing the behavior that we were studying.

Finally, EPs and AHPs in our sample reported lower wiki use for professional purposes (20% and 8%) than reported in a recent review of wiki use in health care. This review identified studies reporting a range of usage rates ranging from 18% for nurse practitioners and physician assistants to 35% for pharmacists, 55% for consultants, and 80% for junior physicians [17,31-34]. Although these differences are possibly due to our small convenience sample, future surveys with larger samples will produce better estimates of current wiki use for professional purposes.

Limitations

Our study has some limitations. First, we had originally planned to use the 2-arm expectancy-value model to measure the influence of indirect constructs (salient beliefs, ie, behavioral beliefs, outcome evaluation, normative beliefs, motivation to comply, control beliefs, and perceived power to influence). However, considering the large number of salient beliefs we retained in our qualitative survey and the fact that the 2-arm expectancy-value model has not shown any advantage over simply using 1 arm of the belief-based measures (only measuring behavioral beliefs, normative beliefs, and control beliefs), we decided to only include items measuring these 3 belief-based measures in our questionnaire [35]. This reduced the number of items in our final questionnaire and likely lowered its administration time, thus reducing participant fatigue and the boredom of answering questions that seem redundant in the 2-arm expectancy model. Shorter questionnaires have been shown to produce more valid information [36].

Second, certain indirect items in our questionnaire lacked temporal stability (eg, control beliefs). This lack of temporal stability might be due to participants changing their assessment of the importance of the different facilitators after 2 weeks, especially if they decided not to watch the video before the retest to save time. Although the retest instructed participants

to watch the video again, we did not ask participants if they actually followed this instruction.

Third, our use of the TPB limits our capability to directly assess the importance of environmental factors such as organizational readiness for change. The use of the Theoretical Domains Framework to inform our theory-based intervention could correct this [37,38].

Finally, our questionnaire does not measure the determinants of contributing to the wiki, in addition to consulting it. By definition, a wiki is a product of its users and remains relevant only if its users continue to update it and create new content. Getting experts and other members of a wide community to contribute to a collaborative writing project is a difficult task and a theory-based approach will be needed to stimulate and promote this behavior [14,39,40]. Unfortunately, time constraints are a major barrier when studying clinician behavior in the ED [41,42]. Thus, questionnaire length limited the number of behaviors we could assess in this study. Several further behaviors will need to be studied in the future, but we chose the one we felt to be the most important (using the wiki). If clinicians do not intend to consult a wiki during their clinical duties, we need to understand the determinants of this behavior before asking them to update and create content.

Future Studies

The next step will be to use these questionnaires in a larger population of ED health care professionals in the Province of Quebec. However, to use this questionnaire in an even broader population across Canada and internationally, our survey instruments (videos and questionnaires) will need to be translated from French to English and other languages and validated using cross-cultural adaptation of the self-report measures [43]. Although we have produced an English version of our questionnaires (Multimedia Appendices 11 and 12), they still need to be validated with a population of English-speaking health care professionals before they can be used in large surveys. Using these questionnaires in multiple settings and countries will help identify the behavioral determinants that a future theory-based intervention should target in order to stimulate the use of wikis promoting best practices in trauma care around the world. Although our questionnaires already contain certain items that are not exclusively relevant to trauma care and EDs and which could serve as a basis for new questionnaires investigating the intention to use wiki-based reminders in other fields of health care, in order to use them to investigate the use of wiki-based reminders in other settings they need to be adapted and validated.

Conclusion

Our newly developed theory-based questionnaire to measure health care professionals' intention to use wiki-based reminders has adequate validity and reliability for use in a large survey. In the long run, this will help develop theory-based interventions for wikis promoting best practices in trauma care.

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Conflicts of Interest

None declared.

Multimedia Appendix 1

Links to YouTube videos presenting the behaviour studied.

[PDF File (Adobe PDF File), 4KB - resprot_v3i3e50_app1.pdf]

Multimedia Appendix 2

Cognitive interview questionnaire (in French).

[PDF File (Adobe PDF File), 7KB - resprot_v3i3e50_app2.pdf]

Multimedia Appendix 3

AHP questionnaire developed during Phase 2.

[PDF File (Adobe PDF File), 206KB - resprot_v3i3e50_app3.pdf]

Multimedia Appendix 4

MD questionnaire developed during Phase 2.

[PDF File (Adobe PDF File), 212KB - resprot_v3i3e50_app4.pdf]

Multimedia Appendix 5

Comments made by focus group participants and changes made to questionnaire.

[PDF File (Adobe PDF File), 11KB - resprot_v3i3e50_app5.pdf]

Multimedia Appendix 6

Analysis of internal consistency and temporal stability for each item in the MD and AHP questionnaires.

[PDF File (Adobe PDF File), 36KB - resprot_v3i3e50_app6.pdf]

Multimedia Appendix 7

Changes made to the EP questionnaire after the two-week test-retest.

[PDF File (Adobe PDF File), 66KB - resprot_v3i3e50_app7.pdf]

Multimedia Appendix 8

Changes made to the AHP questionnaire after the two-week test-retest.

[PDF File (Adobe PDF File), 74KB - resprot_v3i3e50_app8.pdf]

Multimedia Appendix 9

Final AHP questionnaire (French).

[PDF File (Adobe PDF File), 405KB - resprot_v3i3e50_app9.pdf]

Multimedia Appendix 10

Final MD questionnaire (French).

[PDF File (Adobe PDF File), 403KB - resprot_v3i3e50_app10.pdf]

Multimedia Appendix 11

Final AHP questionnaire (English).

[PDF File (Adobe PDF File), 390KB - resprot_v3i3e50_app11.pdf]

Multimedia Appendix 12

Final MD questionnaire (English).

[PDF File (Adobe PDF File), 394KB - resprot_v3i3e50_app12.pdf]

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Abbreviations

AHP: allied health professional
ED: emergency department
EP: emergency physician
ICC: intraclass correlation coefficient
TBI: traumatic brain injury
TPB: Theory of Planned Behavior

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Protocol

Development of a Communication Protocol for Telephone Disclosure of Genetic Test Results for Cancer Predisposition

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Abstract

Background: Dissemination of genetic testing for disease susceptibility, one application of “personalized medicine”, holds the potential to empower patients and providers through informed risk reduction and prevention recommendations. Genetic testing has become a standard practice in cancer prevention for high-risk populations. Heightened consumer awareness of “cancer genes” and genes for other diseases (eg, cardiovascular and Alzheimer’s disease), as well as the burgeoning availability of increasingly complex genomic tests (ie, multi-gene, whole-exome and -genome sequencing), has escalated interest in and demand for genetic risk assessment and the specialists who provide it. Increasing demand is expected to surpass access to genetic specialists. Thus, there is urgent need to develop effective and efficient models of delivery of genetic information that comparably balance the risks and benefits to the current standard of in-person communication.

Objective: The aim of this pilot study was to develop and evaluate a theoretically grounded and rigorously developed protocol for telephone communication of BRCA1/2 (breast cancer) test results that might be generalizable to genetic testing for other hereditary cancer and noncancer syndromes.

Methods: Stakeholder data, health communication literature, and our theoretical model grounded in Self-Regulation Theory of Health Behavior were used to develop a telephone communication protocol for the communication of BRCA1/2 genetic test results. Framework analysis of selected audiotapes of disclosure sessions and stakeholders’ feedback were utilized to evaluate the efficacy and inform refinements to this protocol.

Results: Stakeholder feedback (n=86) and audiotapes (38%, 33/86) of telephone disclosures revealed perceived disadvantages and challenges including environmental factors (eg, non-private environment), patient-related factors (eg, low health literacy), testing-related factors (eg, additional testing needed), and communication factors (eg, no visual cues). Resulting modifications to the communication protocol for BRCA1/2 test results included clarified patient instructions, scheduled appointments, refined visual aids, expanded disclosure checklist items, and enhanced provider training.

Conclusions: Analyses of stakeholders' experiences and audiotapes of telephone disclosure of BRCA1/2 test results informed revisions to communication strategies and a protocol to enhance patient outcomes when utilizing telephone to disclose genetic test results.

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KEYWORDS

genetic testing; test result disclosure; communication; telemedicine; cancer risk assessment; self-regulation theory of health behavior

Introduction

Dissemination of genetic testing for disease susceptibility, one application of "personalized medicine", holds the potential to empower patients and providers with informed risk reduction and prevention recommendations [1,2]. Genetic testing has become a standard practice in cancer prevention, where genetic testing for cancer susceptibility has become routine for high-risk populations, particularly for breast, ovarian, and colon cancer [2-8]. Heightened consumer awareness of "cancer genes" and other disease susceptibility genomic testing (eg, cardiovascular genetics [9], genetic testing for Alzheimer's disease [10], and multi-gene genetic testing for cancer and common diseases [11-14]) has escalated interest in and demand for genetic risk assessment. Increasing demand for predictive genetic testing to inform prevention and medical management of cancer and other diseases is expected to surpass accessibility to genetic specialists [15-17].Thus, the promise of personalized medicine will require innovative delivery models for effective, efficient predictive genetic testing and risk communication.

Given the complexity and limitations of genetic testing for disease susceptibility, pre- and post-test counseling are recommended across a variety of fields (eg, cancer, cardiology, and neurology) to optimize patients' informed consent, understanding of, and adaptive behavioral and psychosocial responses to genetic test results [2,6,11,12,18,19]. Given the complexity of genetic information, the potential for false reassurance, and the potential for psychological distress (eg, persistent anxiety and guilt about the development of cancer in themselves or offspring), communication of genetic test results for cancer susceptibility has traditionally been conducted in-person by health professionals with genetics training [5,20-23]. With increasing demand for genetic testing and time constraints regarding cancer treatment decisions dependent on test results, some genetic counselors are beginning to incorporate telephone disclosure of genetic test results for select patients [24-29]. Some direct-to-consumer companies have incorporated "streamlining" of pre- and post-test counseling protocols [30], offering BRCA1/2 testing, including pre- and post-test counseling entirely by telephone and the Internet. These services are currently commercially available (eg, InformedDNA, Genetic Counseling Services), and some health insurers (eg, Aetna) have partnered with them in the delivery of these services [31-33]. Thus, modifications to traditional genetic service delivery have begun, in the presence of limited data regarding the impact of these changes on patients, providers, and health care systems [31,34]. These changes represent a critical knowledge gap in the translation, implementation, and dissemination of genetic knowledge into effective clinical practice. Concurrently, there is also increasing use of more complex testing, including multi-gene panels evaluating a number of cancer susceptibility loci of varied penetrance, cancer spectrum, and clinical utility [35]. Whole-exome and -genome sequencing add additional complexity with the potential to unveil disease susceptibility beyond the condition of interest [5]. These advances hold great promise to expand the benefits of testing, but in many cases are also associated with greater uncertainty and complexity, presenting additional challenges for providers delivering pre- and post-test counseling [35]. The capacity for traditional counseling models to accommodate these changes is limited. Thus, there is urgent need for theoretically driven studies that evaluate innovations in communication and delivery of cancer genomic advances in real-world clinical settings, which balance the risks and benefits associated with alternatives to in-person communication, in the context of emerging and increasingly complex genomic testing that will address the potential limitations of and inform the adaptive responses to alternative communicative strategies [16,36-38].

We utilized our preliminary data from patients and providers [25], our team's multidisciplinary expertise, and existing literature of genetic counseling, telephone communication in medical consultations, health communication, and health behavior to develop a communication protocol for telephone disclosure of genetic test results [17,21,39-43]. The goal of this pilot study was to develop and evaluate a protocol for telephone communication of clinical BRCA1/2 (breast cancer) genetic test results that might be broadly generalizable. We utilized our theoretical model to inform short-term cognitive (knowledge) and psychological (state anxiety, general anxiety, and depression) outcomes and to identify audiotaped communication sessions for review. Audiotape reviews and direct stakeholder (patient and provider) feedback were utilized to evaluate and inform refinements to our initial telephone communication protocol [34].

Methods

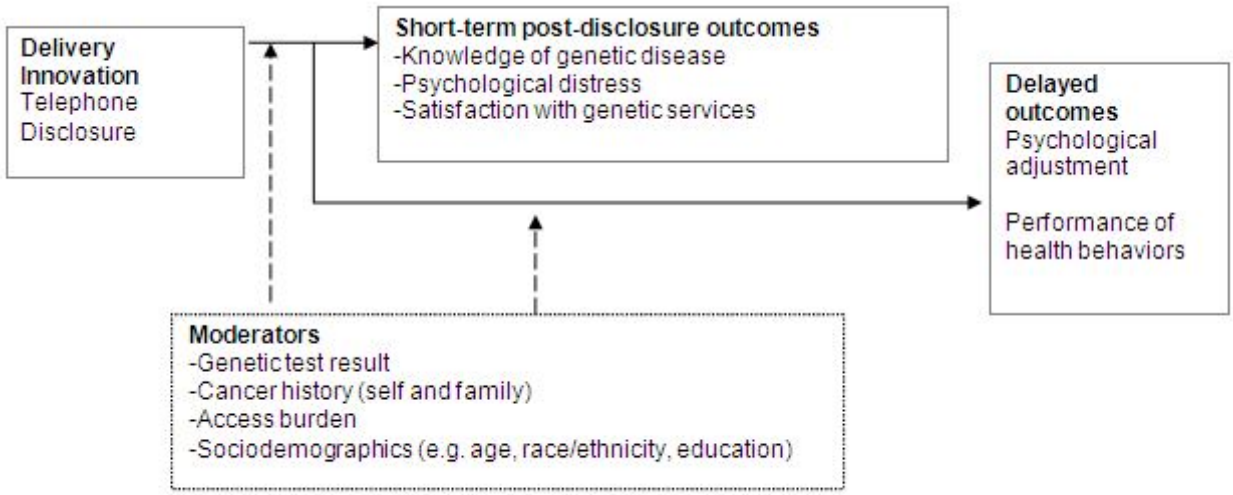
Theoretical Model

This study and related work [25,34,44,45] is informed by our theoretical model to optimize and evaluate the outcomes of innovation to delivery of genetic services (Figure 1). The health benefits of genetic testing for disease susceptibility are expected to be greatest when there are successful interventions to modify disease susceptibility and thus, to improve patient outcomes [11,46,47]. As improved patient outcomes are contingent upon high-risk individuals adopting preventive or promotive health

behaviors, the effectiveness of genetic testing for disease susceptibility is contingent upon successful behavior modification. Our model is grounded in the Self-Regulation Theory of Health Behavior (SRTHB), which has been utilized in descriptive and intervention-based research involving the study of health threats and health behavior [39,48-51]. Simplistically, this theory proposes that the performance of a health behavior is the product of an individual's knowledge and perceptions of the disease threat (eg, genetic risk of disease) and the health behavior (eg, risk reduction behaviors) and the biopsychosocial impact of the health behavior [39,40,49,51-53]. Importantly, the SRTHB emphasizes common-sense representations rather than medical or scientific definitions, and incorporates individual biological, cognitive, emotional, familial, and cultural experiences that might contribute to individual variability in knowledge and perceptions of genetic information, disease etiology, and controllability, and the impact of knowledge and perceptions on risk modification for individuals and their families. It has been proposed that the SRTHB is an ideal framework for considering the psychosocial and behavioral outcomes and thus, the effectiveness of genetic screening for disease susceptibility in the era of personalized medicine [40,42,43]. The SRTHB and associated literature suggests that the performance of risk reduction behaviors and psychosocial adjustment to communication of genetic test results is an iterative process, in which proximal perceptions of and responses to risk information shape more distal behavioral and psychosocial outcomes [39,42,43,54,55]. Thus, the SRTHB suggests an innovative model for the evaluation of both the short-term and long-term responses to novel delivery methods of genetic services, communication of genetic test results, and the impact of that communication on health behaviors (Figure 1).

The literature and our preliminary data support the hypothesis that both short-term cognitive (knowledge and perception) and psychological (state anxiety, general anxiety, and depression) outcomes and longitudinal adjustment and performance of surveillance behaviors, in response to receipt of genetic test results, will be moderated by biological (test result [25,56-58], cancer history [57,59-61]), sociodemographic (eg, race/ethnicity [57,61-63], access burden [25,56]), cognitive, and emotional [26,27,64] factors. Thus, while innovations to delivery of genetic test results might provide equal outcomes in a broad population, there might be subgroups (ie, moderators) for whom this innovation to communication of genetic information is particularly harmful or particularly useful. If the promise of genomics to improve population health is to be realized broadly, it will be critical to understand those factors that have the potential to moderate that outcome for some, and to incorporate that knowledge into the design of future interventions for delivery of genomic information.

Figure 1. Theoretical model to evaluate innovations to delivery of genetic information (guided by the Self-Regulation Model of Health Behavior).



Participants

Institutional Review Board approval was obtained before initiating this study. Participants were recruited through the Fox Chase Cancer Center Risk Assessment Program between September 2009 and July 2010. Eligible patients were over 18 years old, could communicate in English, completed pre-test counseling with a genetic counselor, and elected to proceed with BRCA1/2 testing. Consistent with professional guidelines from the National Society of Genetic Counselors and clinical services at Fox Chase Cancer Center, key components of in-person pre-test counseling included ascertainment of targeted medical and family history, assessment of cancer risk, education on cancer genetics, discussion of appropriate genetic testing options, and informed consent for genetic testing [19,65]. Eligible patients were approached by research staff at completion of pre-test counseling offering the opportunity to participate in a study receiving their BRCA1/2 test results by telephone and returning for in-person medical management recommendations. Written informed consent from patients was obtained prior to conducting audiotaped telephone disclosure sessions. Providers also signed informed consent and completed brief surveys assessing their perception of how participants perceived their disclosure sessions.

Telephone Disclosure Communication Protocol

We utilized our preliminary data from patients and providers [25], our team's multidisciplinary expertise, existing literature of genetic counseling, health communication, telephone communication in medical consultation, and our theoretical model grounded in SRTHB to develop a protocol for telephone disclosure of genetic test results [5,17,21,39-43,66,67]. The key components, although tailored for this study to the disclosure of BRCA1/2 testing results by telephone, were designed to be broadly adaptable to communication of genetic/genomic test results, hereditary risk, and risk reduction strategies for other heritable diseases. Key components include (Table 1): "Visual Aids", "Standardized Communication Topics", and "Provider Probes"; "standard probes" are used intermittently throughout the session to assess patient understanding (eg, "What questions do you have for me before we go on?") and affect (eg, "How are you feeling now that you know…?"). At the conclusion of the session, a "Teach Back" probe (eg, "Please tell me in your own words your understanding of your genetic test results and what those results mean for you") is used to assess comprehension [67]; "situational probes" are used as needed (eg, "It sounds like this might not be the best time for us to talk. Is there another time that would work better for you?") in contexts where the situation might present challenges to optimal outcomes. Other key components include "Provider Training": in-person training was provided to genetic counselor (GC) providers to optimize the translation and implementation from in-person to telephone disclosure; and "Quality Assurance and Protocol" evaluation where all telephone disclosure sessions were audiotaped. Sessions' meeting criteria (see Statistical Analyses) were reviewed by research staff in tandem with disclosure checklists completed by the genetic counselors to (1) ensure inclusion of 12 key components of disclosure [34], and (2) inform potential refinements to the communication protocol. Additionally, all participants who completed telephone disclosure were asked to return for an in-person follow-up appointment with a physician to address any remaining questions and to discuss and implement medical recommendations for cancer screening and/or risk reduction strategies (ie, prophylactic surgery and/or chemoprevention) [34].

Table 1. Key components of telephone communication protocol for BRCA1/2 testing.

Components		
Visual Aids		
		Pedigree
		Etiology
		Heritability
		Associated cancer risks
		Sample genetic test results
		Risk reduction options
Standardized Communication Topics		
		Confirm patient's identity
		Introduce all participants
		Assess adequacy of hearing and access to Visual Aids
		Affirm session purpose and patient's desire for results
		Provide test results, interpretation, and implications
Provider Probes		
	Standard - Evaluate patient:	
		Understanding
		Emotional response
	Situational - Address:	
		Session distractions
		Patient interruptive
		Patient emotional
		Patient disengaged
		Others present in session reactions/needs
	Teach Back:	
		Evaluate patient understanding at conclusion
Provider Training		
	In-person	
		Training manual
		Challenges to telephone communication
		Mock telephone disclosure w/Individualized feedback
Quality Assurance		
	Evaluation	
		Innovations in delivery of genetic/genomic information
		Inclusion of key components
In-Person Follow-up		
	Appointment with physician	
		Address remaining questions
		Discuss and implement medical management recommendations

Measures

Overview

Patients completed quantitative assessments of knowledge [68-70], psychological distress (state anxiety) [71], psychological adjustment (general anxiety and depression) [72], and satisfaction with genetic services [73-75] previously reported [44] within 3-5 days after their pre-test counseling session (baseline) and telephone disclosure. Participants completed 7 selected items consistent with the National Institute of Health National Center for Human Genome Research Cancer Genetics Consortium Knowledge scale and utilized in prior research [69], including items evaluating mechanism of cancer inheritance (1 item), and the meaning of positive (3 items) and negative (3 items) results. Internal consistency in this study was good (mean alpha=.72).

Psychological Distress

State anxiety was measured with the 20-item State Inventory of the State-Trait Anxiety Inventory (STAI), which is a sensitive indicator of transient or situational changes in anxiety (test-retest reliability: r=16 - .62) experienced by patients in response to stressful procedures or life events [71] and is frequently used to assess the short-term response to the receipt of a genetic test result [57,76]. Internal consistency in this study was high (mean alpha=.95). General anxiety and depression were assessed with the 7-item Hospital Anxiety and Depression Scale (HADS) anxiety and depression subscales, which have been utilized in the general population and a wide range of medical patients, including those with cancer [72,77]. Internal consistency in this study was good for both the anxiety and depression subscales (mean alpha=.88, .78, respectively) [34].

Satisfaction With Genetic Services

Satisfaction with health communication was measured with a 9-item scale, reflective of constructs identified in prior qualitative, quantitative, and comparative research evaluating participants' perceptions of their genetic counseling and testing experience, including cognitive, affective, and time/attention items [73-75]. Internal consistency in this study was good (mean alpha=.73) [34].

Opinions and Experiences Regarding Telephone Disclosure Post-Disclosure Only

Patients and providers also completed parallel open-ended questions after telephone disclosure to elicit patient experiences (3 items), GC experiences (3 items), and GC perceptions of patient experiences (3 items) with, and suggestions for improving their telephone disclosure session [34].

Statistical Analyses

Changes in pre-test and 3-5 days post telephone disclosure scores were calculated for each participant and each construct, which are reported separately [34]. Pre-defined criteria were developed to select telephone disclosure sessions for review to inform modifications to the communication protocol [34]. These included (1) all positive results (n=9), (2) discordance between patient reported satisfaction, perceived understanding/ emotional response, and provider perceptions of patient satisfaction/ understanding/ emotional responses; discordance was defined as a difference of >2 points between patient and provider for any individual item (n=12), (3) provider request (n=1), (4) decline in knowledge in the lowest 10 percentile; these included a decline in knowledge of >3.3 points (n=8), and (5) increase in state anxiety, general anxiety, or depression in highest 10 percentile (n=21). These included increases in state anxiety scores >11.3 points, HADS-anxiety, or HADS-depression subscale scores >3 points. These selection criteria resulted in review of 38% (33/86) of recorded telephone disclosures. Participants selected for review did not differ statistically from those not selected for review on race, age, education, test result, cancer history, treatment decision, and known familial mutation.

Framework analysis was utilized to analyze open-ended responses regarding patient and GC reported advantages, disadvantages, and recommended modifications (eg, "What did you/your patient like/dislike about receiving genetic test results by telephone? What changes would you recommend?") [36,57,78-80]. The intent of this analysis was to identify themes that might not have been represented in quantitative surveys, informing modifications to the intervention from both patient and provider perspectives. Two investigators, a clinical health psychologist trained in health outcomes research, and a medical oncologist, each with extensive clinical experience in the delivery of hereditary cancer risk information, independently reviewed responses utilizing thematic analysis to record primary and secondary themes for each open-ended item.

Results

Participant Characteristics

A total of 167 eligible subjects were approached for participation. Of the 167 eligible subjects, 100 (59.9%) agreed to participate, provided informed consent, and completed the baseline assessment, and 95 proceeded with BRCA1/2 testing [34]. A total of 86 participants completed both baseline and post-telephone disclosure surveys, and participant characteristics are described in Table 2.

All participants were women, 10% (9/86) were non-white, 59% (51/86) had a personal history of cancer, and 50% (43/86) had a college degree or graduate education. The majority of participants (83%, 71/86) received an uninformative negative result (ie, negative BRCA1/2 result with no known clinically significant mutation in the family). Nine women received a positive result (ie, clinically significant BRCA1/2 mutation), 4 received a true negative result (ie, negative for a known clinically significant mutation in the family), and 2 received a variant of uncertain significance. Provider checklists revealed high fidelity to communication topics [34].

Table 2. Participant characteristics (participants who completed both pre and post disclosure assessments) (n=86).

Characteristic		n (%)
Age, median (range)		49 (24-73)
Race		
	White	77 (90)
	Black / African American	6 (7)
	Asian	3 (3)
Education		
	High school only	13 (15)
	Some college / vocational	30 (35)
	College degree	21 (24)
	Graduate degree	22 (26)
Marital status: married/domestic partnership		54 (62)
History of cancer		51 (59)
Treatment decision[a]		19 (22)
Known mutation in family		6 (7)
Genetic test (BRCA 1/2) result		
	Indeterminate (uninformative negative)	71 (83)
	Positive	9 (10)
	True negative	4 (5)
	Variant of unknown significance	2 (2)

[a]Defined as individuals who had not received definitive surgical treatment for their breast cancer at the time of initial counseling.

Patient and Provider Open-Ended Responses

Patients and providers identified several advantages for patients to telephone disclosure of genetic test results, including patient conveniences, setting, and timing. Patient and provider reported advantages for patients did not differ. Providers also identified advantages of telephone disclosure for genetic counselors, including setting, scheduling, and efficiency (Table 3). Most patients and genetic counselors reported no disadvantages to telephone receipt of test results for patients. Primary themes reported by patients and genetic counselors were communication, delivery, and patient specific challenges (Table 3). Providers reported similar disadvantages for genetic counselors, which also focused on communication and delivery challenges, as well as patient specific factors, such as providing a positive or variant of uncertain significance (VUS) result.

Table 3. Advantages and disadvantages to telephone disclosure: open-ended survey responses from patients and providers.[a]

Advantages / disadvantages
Patient advantages reported by patients and genetic counselors/providers
Patient conveniences, eg, less travel, lost work time, need for dependent care
Setting, eg, being in a comfortable environment, having a support person available
Timing, eg, receiving results prior to MD appointment
Provider advantages reported by genetic counselors/providers
Scheduling, eg, greater availability and flexibility for scheduling patient appointments
Setting, eg, having access to resources in provider's office during appointment
Efficiency, eg, no travel to and wait in clinic
Patient disadvantages reported by patients and genetic counselors/providers
Communication, eg, not being able to see the genetic counselor for nonverbal communication
Delivery, eg, interruptions (workplace, children in the home)
Patient-specific factors, eg, receiving positive or uncertain results
Provider disadvantages reported by genetic counselors/providers
Communication, eg, not being able to see the patient for nonverbal communication
Delivery, eg, interruptions (workplace, children in the home)
Patient-specific factors, eg, delivering positive or uncertain results

[a]From 86 encounters (n=86 patients, n=4 providers).

Modifications to the Communication Protocol

Modifications to the original communication protocol were informed by the audiotape review (33/86, 38%) as described above, open-ended survey responses from patients (n=86) and providers (n=4) on 86 disclosure sessions, our theoretical model, and the literature, and developed by a clinical health psychologist. Primary modifications are summarized in Table 4 and included GC clarification of patient instructions for telephone disclosure during pre-test counseling (eg, reviewing visual aids), and appointments for telephone disclosure scheduled to allow patients to plan to be in a private, comfortable place where they would be free of interruptions. Scheduling the disclosure session also allowed patients to include significant others in the session. Additionally, it had the potential to reduce anticipatory anxiety and eliminate the frustration of "playing phone tag". Other modifications included refinement of visual aids, including more clearly labeling mock test report forms to avoid patient confusion, and more detailed telephone disclosure checklists were created for GCs to ensure the communication protocol's coverage of all critical elements of disclosure (eg, Genetic Information Nondiscrimination Act, or GINA), to enhance fidelity to the protocol (eg, reminder that session will be audiotaped), and to facilitate use of verbal knowledge and affective probes, critical to the assessment of understanding and emotional response in the context of telephone communication where nonverbal cues are not available for this purpose. Another modification included GC training to facilitate identification and management of situations that might present unique challenges in the context of telephone communication. Situations addressed included identifying (1) patients' biopsychosocial risk factors prior to disclosure session (eg, personal/familial cancer history; baseline high anxiety or low health literacy; uninformative test results, or need for additional testing), (2) signs of potentially poorer affective and cognitive responses during result disclosure (eg, silence, pressured speech, incongruous affect), and (3) need for session management (eg, interruptions), in the absence of the visual information providers are accustomed to incorporating into their assessments of patients in the conduct of in-person sessions. Training included, in addition to regular utilization of affective and knowledge probes and the "teach-back", increased attention and response to auditory cues in patients' speech, eg, rate, pitch, volume, amount, type and congruity; and utilization of situational probes, eg, "Did I catch you at an inconvenient time…", "Would it be ok to check in now with your (spouse, mother, sister, etc?)", "Is this the information you were expecting, or how can I better help you meet your needs?"

Table 4. Modifications to communication protocol resulting from stakeholder (patients and genetic counselors) surveys (n=86) and session tape reviews (n=33).[a]

Modifications to the communication protocol	Reason for tape review	Reviewer observations	Patient comments	GC[b] comments
1. Clarified telephone disclosure (TD) instructions in pre-test counseling			-Visual aid: read in advance and have for session	-Visual aid: read in advance and have for session
			-Schedule sufficient time for session and processing	-Schedule sufficient time for session and processing
			-Have support person	-Have support person
2. Scheduled TD appointments	-Increase in anxiety or depression or decline in knowledge	Session occurred in non-private environment without visual aids	-Session was disrupted (eg, workplace, childcare)	-Session was disrupted (eg, workplace, childcare)
			-Session disrupted other activities	-Difficult to reach patient ("phone tag")
3. Refined visual aids			Visual aids confusing	Visual aids confusing
4. Improved disclosure checklist:	-Increase in anxiety	-Some elements of disclosure checklist omitted		Who else is on call/present?
-Enhanced formatting				
-Included information on GINA[c]		-Patient concerned about genetic discrimination		
5a. GC training:	-Increase in anxiety or depression	-High baseline anxiety	-Communication challenging without visual cues	Interpreting patient affective response and providing emotional support challenging without visual cues
-Recognizing signs of negative affect in the absence of visual cues	-Patient/GC discordance	-Inaccurate expectations		
-Effective use of affective and situational probes				
-Need to pace session to meet patient needs				
-Identifying risk factors for negative affective response	-Positive test result	-Personal history of cancer	-Need to be prepared for all results	
		-Treatment decision pending		
		-Family history (uninformative or extensive cancer)		
		-Need for additional tests (self or family members)		
5b. GC training:	-Decline in knowledge	Low health literacy		Interpreting patient cognitive response and providing remediation challenging without visual cues
-Identifying risk factors for confusion				
-Recognizing signs of confusion				
-Techniques to improve patient comprehension	-VUS[d] test result			
-Effective use of knowledge and situational probes and "teach back" to assess understanding				
5c. GC training:	-Decline in knowledge			-Responding to challenging patients/situations

Modifications to the communication protocol	Reason for tape review	Reviewer observations	Patient comments	GC[b] comments
-Effective use of situational probes to control situation				-Controlling the session
6. Conduct larger trial to evaluate outcomes for potentially vulnerable subgroups	-Positive test result	-Need for additional testing (self or family members)	TD might be more challenging in some situations (eg, Positive test result, need for additional testing, poor understanding after pre-test counseling, psychological factors)	TD might be more challenging in some situations (eg, Psychosocial comorbidities, English as a second language, pending treatment decisions, personal cancer history)
	-VUS test result	-Personal history of cancer		
		-Family history of cancer		
	-Increase in anxiety or depression	-Uninformative negative result		
		-Insurance issues		

[a]Based on a priori review criteria

[b]GC: genetic counselor

[c]GINA: Genetic Information Nondiscrimination Act

[d]VUS: variant of uncertain significance

Discussion

Principal Findings

The goal of this pilot study was to evaluate stakeholder (patient and provider) experiences with our telephone disclosure protocol, grounded in our theoretical model, to inform refinements to our initial protocol for the delivery of genetic test results and to obtain preliminary outcome data for future research. To our knowledge, this is the first study to describe the development and refinement of a telephone communication protocol for the disclosure of genetic test results informed by provider and patient feedback, as well as systematic review of communication sessions grounded in a theoretical model of health behavior [42,43]. This study provides a standardized model for optimizing telephone communication that will allow future studies to implement and evaluate the efficacy, risks, and benefits of telephone disclosure of genetic test results. This protocol includes a number of innovations made in response to our findings, supported by our theoretical model and the literature regarding inclusion of telephone communication into primary care. These include the need for specific training for providers to learn compensatory techniques for telephone communication [34,66] (eg, the need to implement more verbal probes in the absence of nonverbal cues) [26,27,34,64,81,82], and have suggested that telephone disclosure might provide equal or improved outcomes when compared to in-person disclosure [26,27,64,83]. This is consistent with our data [34,84] and primary care literature suggesting that telephone might be better suited to encounters following an initial in-person consultation [66]. Given an increased demand for genetic counseling services and the increasing availability of many new modes of genetic and genomic testing (eg, multi-gene, germline, tumor, and pharmacogenomic testing), our study has focused on reconciling provider and patient perspectives on telephone communication to formulate an effective and efficient telephone communication protocol delivery of genetic test results.

The results of our study indicate that patients and providers identify both advantages and disadvantages of telephone disclosure of genetic test results (Table 3). Advantages, highlighted by both patients and providers, included the convenience, scheduling, setting, timing, and efficiency of telephone disclosure. Perceived disadvantages to telephone communication, associated with the loss of nonverbal communication, included the possibility that some subgroups of patients (eg, positive test results, pre-existing psychosocial or cognitive comorbidities) might be more vulnerable to poorer outcomes with telephone communication of genetic test results. In the absence of a randomized trial, it is not possible to know if patients in these subgroups might also have fared differently from others had they received their results in person. Patient and provider feedback and review of disclosure session audiotapes suggested many modifications that are expected to optimize communication of genetic test results by telephone. The results of our study illustrate the challenges in assessing patients' emotional response and understanding via telephone and providers' response to those patient needs. They emphasize the importance of identifying and developing the training needed to establish skills necessary for novel modalities of communication of genetic/genomic information.

To address these challenges, provider training, including additional verbal probes and procedures to assess patients' psychological response and understanding, as well as situations unique to non-in person communication, must be incorporated into counseling to ensure effective communication of health information via telephone. Professional or educational training for alternative counseling models that incorporate these probes and procedures could prove invaluable for future counseling for both novice and experienced genetic counselors. Evolving genetic technologies have the potential to expand the reach of

genetic testing. Realizing the associated health gains will require a workforce of genetic counselors with training in telephone and other nontraditional models of communication of genetic information. Research has shown that effective communication between providers and patients can be achieved through these technologies and might result in improved outcomes [85]. Future studies must focus on evaluating the potential disadvantages of telephone disclosure for vulnerable subgroups in randomized clinical trials.

In this paper, we present a model for adapting the standard genetic counseling model for innovations in the delivery of genetic medicine. Our model includes stakeholder (patient and provider) input, as well as systematic review of audiotapes of pilot sessions of telephone disclosure of genetic testing results and is theoretically informed. We expect this model will be broadly applicable to adaptions of counseling for other innovations in the delivery of genetic/genomic test results. For example, we are currently employing adaptations of this model to study the potential to increase access by utilizing telemedicine to deliver genetic counseling services to community practices where genetic counselors are not available [45], to inform modifications to genetic counseling and delivery of multi-gene testing for cancer susceptibility for patients with suspected hereditary breast and ovarian cancer (HBOC) and other hereditary cancer syndromes, including hematological and gastrointestinal syndromes, and to inform prescriptive decisions for providers. We expect this model to be broadly generalizable to evolving contexts in genetics and medicine as innovations develop.

Limitations

We acknowledge several limitations to this study. Despite the recruitment of a clinical population, we had relatively low representations of some subgroups (eg, ethnic minorities and those of low socioeconomic status). Thus, our findings may not be generalizable to a more diverse population. Our on-going multi-center randomized study aims to increase representation of these subgroups. It will also inform how cognitive and affective responses might differ between patients receiving in-person and telephone communication of genetic test results.

Conclusions

Providers delivering and patients receiving genetic test results by telephone identified advantages to telephone disclosure (eg, perceived convenience for patients and providers) and disadvantages (eg, loss of non-verbal cues to assess and communicate emotional responses and understanding). Review of stakeholder reported experiences and audiotapes of telephone disclosures lead to a number of communication strategy and protocol revisions to enhance patients' cognitive, affective, and behavioral responses to genetic test results in the context of telephone disclosure of genetic test results.

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Conflicts of Interest

None declared.

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Abbreviations

GC: genetic counselor
GINA: Genetic Information Nondiscrimination Act
HADS: Hospital Anxiety Depression Scale
SRTHS: Self-Regulation Theory of Health Behavior
STAI: State-Trait Anxiety Inventory
TD: telephone disclosure
VUS: variant of uncertain significance

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Original Paper

Active Patient Participation in the Development of an Online Intervention

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Abstract

Background: An important and challenging part of living with cancer relates to the repeated visits to the hospital. Since how patients cope between these post-diagnostic visits depends partly on the information and support received from their physician during the visits, it is important to make the most of them. Recent findings reinforce the importance of training not only the health care professionals in communication skills, but providing patients with support in communication as well. Delivering such supportive interventions online can have potential benefits in terms of accessibility, cost-effectiveness, and ability to tailor information to personal needs. However, problems with attrition (dropout, non-usage) during the test phase and poor uptake after implementation are frequently reported. The marginal level of engagement of the patient as end user seems to play a role in this. Therefore, recent research suggests integrating theory-based development methods with methods that promote involvement of the patient at an early stage. This paper describes a participatory protocol, used to let patients guide a theory-informed development process.

Objective: The objective of this project was to apply a bottom-up inspired procedure to develop a patient-centered intervention with corresponding evaluation and implementation plan.

Methods: The applied development protocol was based on the intervention mapping framework, combined with patient participatory methods that were inspired by the participation ladder and user-centred design methods.

Results: The applied protocol led to a self-directed online communication intervention aimed at helping patients gain control during their communications with health care professionals. It also led to an evaluation plan and an implementation plan. The protocol enabled the continuous involvement of patient research partners and the partial involvement of patient service users, which led to valuable insights and improvements.

Conclusions: The applied protocol realized patient participation on different levels throughout the entire project. Early involvement, involvement on different levels, and flexibility in terms of planning and setup seem to be preconditions to creating a bottom-up inspired development procedure with (seriously ill) patients. Further research is necessary to find out if a more patient-centered approach improves the implementation and uptake of eHealth interventions.

Trial Registration: Netherlands National Trial Register ID number: NTR3779; http://www.trialregister.nl/trialreg/admin/rctview.asp?TC=3779 (Archived by WebCite at http://www.webcitation.org/6TdfALKxV).

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KEYWORDS

communication; malignant lymphoma; online intervention; self-help application; patient participation; intervention development

Introduction

An important and challenging part of living with cancer concerns the repeated visits to the hospital. These visits are important as they monitor the development of the disease and set the stage for how to cope with life until the next consultation. Since how patients cope between these post-diagnostic visits depends partly on the information and support received from the health care professionals (HCPs) (eg, specialists, nurses) during the visits, it is important to get the most out of them. Many training programs are designed to improve HCPs' communication skills, which may facilitate patient engagement in the medical dialogue. However, cancer patients ascribe many barriers in medical communication to their own attributes, such as a lack of communication skills, and interfering emotions and beliefs [1,2]. These findings reinforce the importance of training not only HCPs in communication skills, but providing patients with support in communication as well. Epstein and Street (2007) have stressed the need for developing specific types of interventions to support cancer patients, such as in-person coaching, interactive computer programs, videos of role models, and question prompt sheets [3].

Such interventions can be especially efficient when delivered online. The content and type of online interventions can be computer tailored to patients' preferences and needs and they can be accessible any time and any place in a cost-effective way [4]. With regard to knowledge and skill building, the effects of online interventions for patients seem to be equivalent to traditional medical education methods (eg, a brochure or human-delivered intervention) [5,6]. Despite these potential benefits, problems with attrition (dropout, non-usage) during the test phase and poor uptake after implementation are frequently reported [7-9]. According to Eysenbach, characteristics related to the participants, the intervention, and the study design influence the usage and adoption success of online interventions [10].

The technology- and expert-driven development methods (top-down) are indicated as possible causes for attrition and adoption problems [11]. These imply a marginal level of engagement of the involved end-users (especially patients). Therefore, recent research suggests integrating methods that promote involvement of the patient at an early stage (bottom-up) with theory-based intervention development methods [11,12]. Patient participation is frequently referred to, the potential benefits are widely accepted, and there is a clear urge for more patient involvement [13]. However, the actual operationalization, that is, how and when (seriously ill) patients are involved, is rarely reported [14,15]. It often seems a more symbolic statement or it is used to describe the participation of patients in health programs. This differs from patients' active involvement in the organization, goal setting, planning, and execution of interventions [16].

Considering the fact that the contribution of patients in oncology consultations is often limited [3,17,18] and that patients ascribe many communication barriers to personal attributes [1,2], the PatientTIME project was set up (Patients Talk In Medical Encounters). In this project, an online intervention is developed, tested, and implemented that aims to teach patients to take more control during their consultations. The project aims to realize this with a bottom-up inspired approach, which implies the involvement of seriously ill patients throughout the entire project. The initiation of the project was triggered by a specific request for support in communication with HCPs, expressed by a group of patients diagnosed with malignant lymphoma. Lymphoma patients often face long, intense treatment periods and/or monitoring periods under specialist care, which involve many hospital visits. Apparently, despite the (mainly paper-based) information available for this group, patients with malignant lymphoma experience difficulties in communicating their own agenda and needs to their HCP.

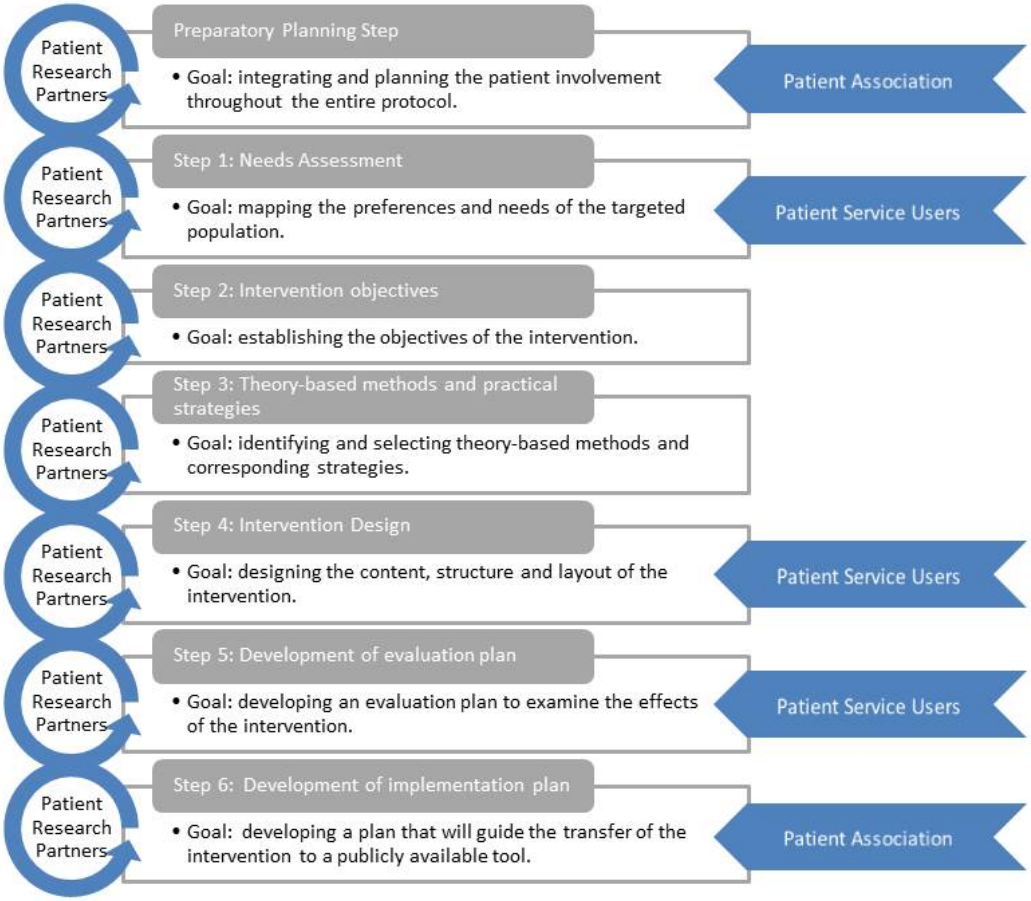
This paper outlines the patient participatory approach used to develop an online intervention with corresponding evaluation and implementation plan. The goal of this paper is to share the applied protocol, the use of the protocol in the PatientTIME project, and our lessons learned in the attempt to create a bottom-up inspired intervention.

Methods

Outline

A stepwise protocol (Figure 1) was applied to develop the intervention with corresponding evaluation and implementation plan. For each step, goals were set and the procedure to involve patients was planned in advance. The Intervention Mapping (IM) framework was used as theoretical backbone of the protocol. Aiming at a patient-driven development protocol, practical patient participatory methods were integrated in the theoretical IM framework and used to inspire when and how patients could be involved.

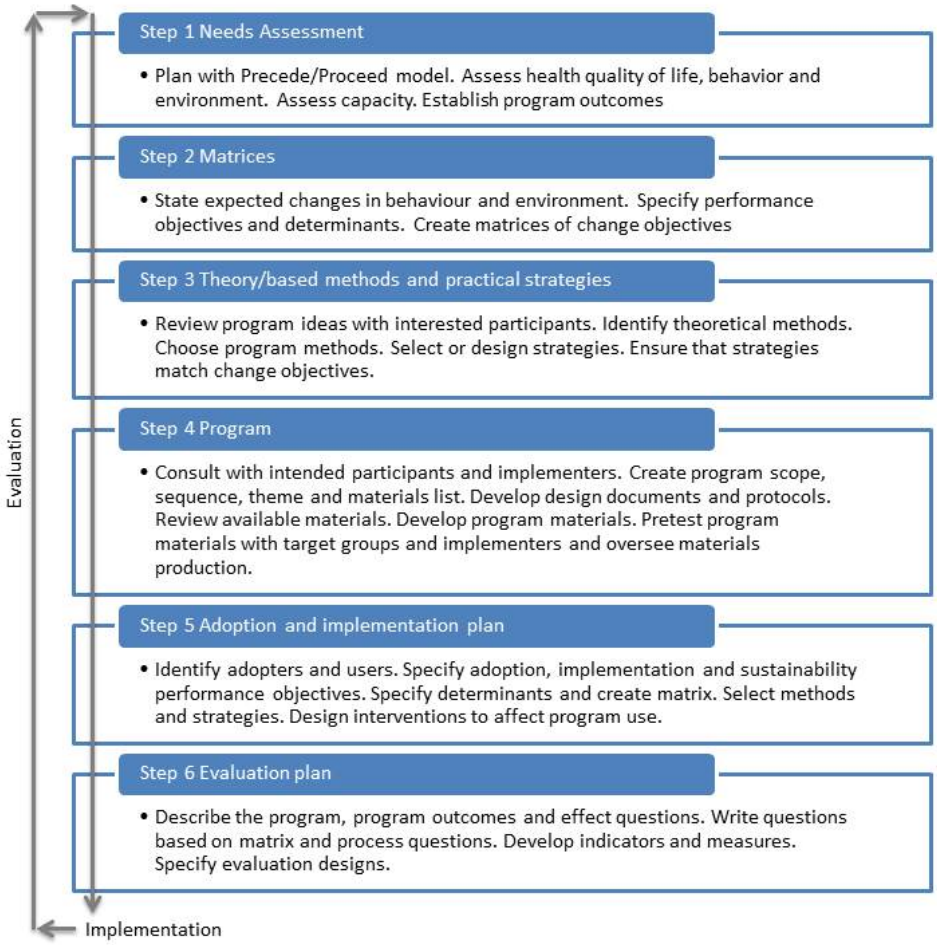
Figure 1. Stepwise protocol.



Intervention Mapping as a Theory-Based Guideline

The IM framework systematically guides the planning and decision-making process in health promoting programs [19]. It comprises six steps in the process toward the development of a theory-driven and evidence-based intervention (Figure 2). The outcome of each step guides the next step. The IM framework has already been used successfully in developing a range of eHealth programs [20-23]. The IM framework was chosen as a guideline because it links decisions, final materials, and activities to theory. A preparatory step was added to the IM framework to plan and prepare the patient participation throughout the entire protocol.

Figure 2. Intervention Mapping framework.



Patient Participatory Methods

The way patients were involved in the applied protocol was inspired by the concept of participation ladders. Different participation ladders describe the idea of involving participants in varying degrees [24-27]. Definitions of these degrees vary, but they all describe a stepwise scheme from no participation (eg, patients participate but have no understanding of the project; they get information but there is no dialogue) to the highest possible level of participation (participants directly collaborate with the stakeholders; have an agenda-setting, initiating role). This concept inspired us to involve patients on different levels and we operationalized this by (1) setting up a close collaboration with the patient association for malignant lymphoma (Hematon), (2) recruiting patients as research partners, and (3) planning the involvement of patient service users. Hematon informs and supports patients and champions patient interests. Patient research partners are involved throughout an entire project and they are equal partners in a working group. Patient service users are involved on different levels, in different parts of the project.

User-centered design (UCD) was used as a guide to realize patient participation in the different protocol steps. UCD is defined by Preece et al (2002) as “an approach, which views knowledge about users and their involvement in the design process as a central concern”. The challenge of UCD is to map the needs, behavior, actions, and abilities of the end user and let this information influence how the intervention takes shape. The context mapping method (Step 1) and the usability tests (Step 4) were inspired by UCD thinking.

Patient Recruitment

All participating patients were adults diagnosed with malignant lymphoma and they all voluntarily signed up to contribute to the project. They were recruited via social media, online newsletters, advertisements on Hematon’s website, regional and national patient conferences, and leaflets in hospital waiting rooms. To recruit patient research partners, Hematon informed several of their active volunteers (patients) who had experience in information and communication technology (ICT) development and with supporting fellow patients.

Project Management

A multidisciplinary working group consisting of researchers, HCPs, and a patient research partner was responsible for the daily coordination of the project. The working group collaborated with physicians, nurse practitioners, patients, user-interaction designers, software developers, and representatives of Hematon. Final decisions regarding the protocol were reached through discussions in the working group. Decisions related to the implementation plan were made in consultation with Hematon.

Results

Overview

The intervention development protocol resulted in three products: a self-directed online communication tool, a corresponding evaluation plan, and an implementation plan. The goal of the intervention is to help patients gain more control in the communications with their HCPs. Patients can access the intervention before each hospital visit. The information is provided via an algorithm computer-tailored to the patient's self-assessed, momentary efficacy for communication with their HCP, to whether he or she attends the HCP alone or with a companion, and to the stage of treatment. The central information consists of short video clips of simulated consultations that model adequate communication behavior. Additionally, the intervention includes an open question prompt sheet (QPS), a reminder system linked to a list of planned hospital visit dates, and an option to store and play back audio recordings of the consultation (see Multimedia Appendix 1). The evaluation plan comprises a randomized controlled trial (RCT) protocol, in which the effects of the intervention on the patients' perceived efficacy are measured in a trial setting. In the implementation plan, the conditions are built to transfer the evaluated intervention to a publicly available tool. The following paragraphs outline how the patient participatory protocol was used to develop these three products.

Patient Participation Planning (Preparatory Step)

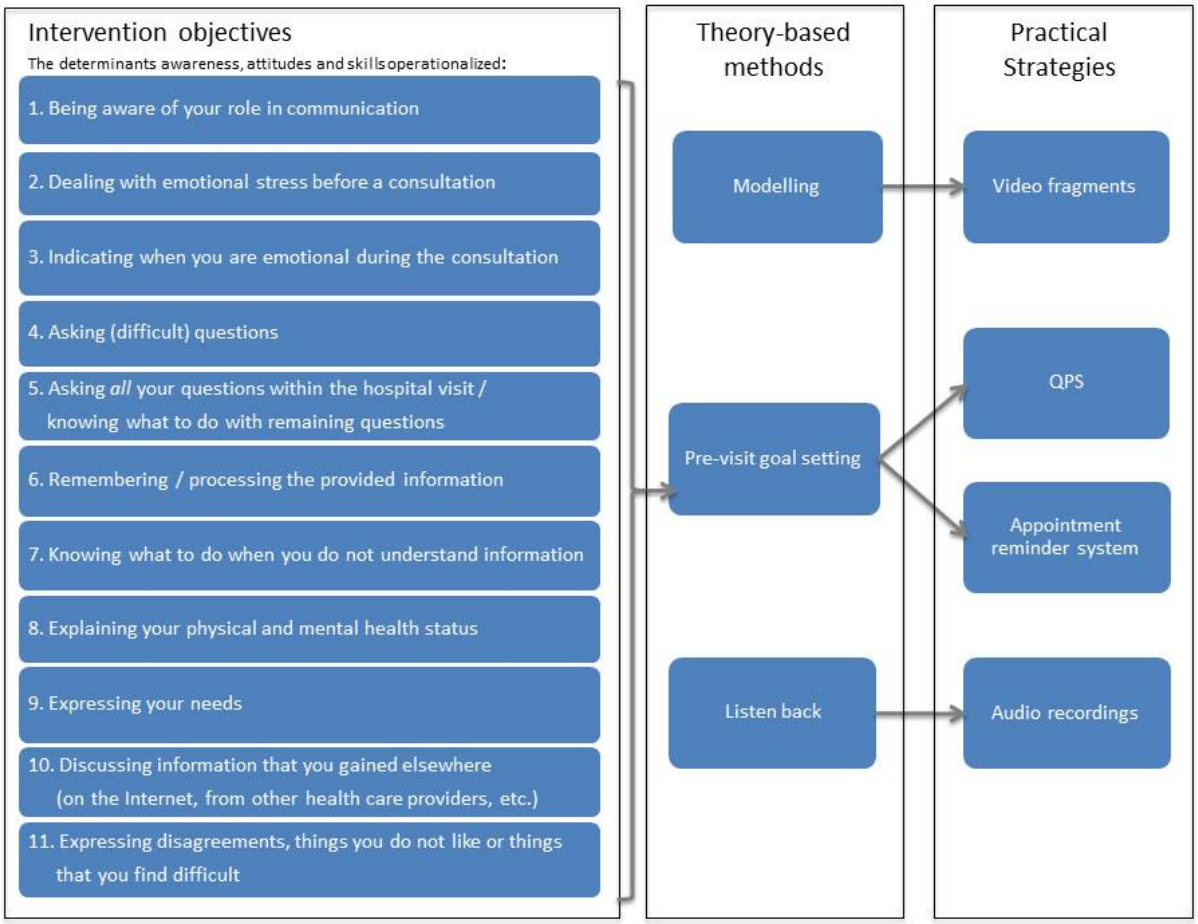
The goal of this preparatory step was to integrate and plan the patient involvement throughout the entire protocol. This resulted in the recruitment of two patients as research partners. They both had been active in supporting fellow patients and therefore they had built a rich body of knowledge about the different aspects of having malignant lymphoma. Additionally they both had a relevant professional background in ICT (Web-development, system design, research and development). One research partner (HG) became part of the working group. The second research partner was consulted on a more irregular basis. The research partners were directly involved in the planning of the PatientTIME project and in the decision-making processes in each protocol step. This involvement approach aligns with the upper steps of the participation ladder as they had an initiating and agenda-setting role and they worked directly with the other stakeholders. Additionally, patient service users were invited to participate in the needs assessment (Step 1), intervention design (Step 4), and the evaluation (Step 5). Moreover, their input was used to inspire the other protocol steps. Last, representatives of Hematon were consulted to explore the possibilities for implementing the intervention after the research project has ended (Step 6) and how we could use their network to keep in close contact with patients.

Needs Assessment (Step 1)

The goal of the needs assessment step was to map the patient-perceived barriers and facilitators in communication with HCPs and to learn from patients' experiences. A qualitative two-step method was applied, inspired by user-centered design thinking. The applied method is derived from the context mapping framework, used by product developers and user interaction researchers to gain insight into the needs of prospective users of new products [28]. Details of this study are described elsewhere [2]. In short, patients completed a set of assignments about their experiences with medical consultations, aiming to trigger them to verbalize and reflect on experiences, preferences, and needs without the presence of researchers or other patients. This so-called sensitizing process is supposed to enhance the quality and quantity of patients' contributions in later (group) interviews [29]. Subsequently, these patients and their spouses shared their experiences during semi-structured (group) interviews, which were audio-recorded. Before conducting this needs assessment, a patient research partner reflected on the study design and the formulated questions. According to his feedback, the introduction was changed to further clarify the goals of the study, more and other examples were added to illustrate the questions, and subtle changes were made to the formulation of questions (eg, avoiding medical jargon, less formal style). A total of 37 patient service users (28 patients and 9 spouses) contributed to this needs assessment. They were open, willing, and motivated to share their experiences and they all had experienced difficulties in communication during consultations. Many communication barriers were ascribed to their own attributes (eg, emotions, skills, and beliefs).

The expressed barriers were analyzed, clustered, and translated into a list of intervention objectives (Step 2) and used as a basis for the central information of the intervention (Step 4). For example, patients did not want to be bothersome and therefore they found it hard to ask (all of) their questions and to express details about their physical and/or mental health status. This information was used to develop information about how to request attention for your prepared questions (Figure 3, Objective 4) and about the importance of expressing your physical complaints and worries (Figure 3, Objective 8). Participants also reported that their communication attitude and skills changed over time, and so did their perceived barriers and facilitators. This finding stressed the need to inquire about the patient's needs before every hospital visit and tailor information accordingly.

Figure 3. Intervention objectives, theory-based methods, and practical strategies.



Intervention Objectives, Theory-Based Methods, Practical Strategies (Steps 2 and 3)

The goal of the second and third step was to establish the objectives of the intervention by specifying what would change as a result of the intervention. The overall aim of the intervention is to support patients in effective communication by creating awareness about the role they can play and the benefits they can gain from participating, as well as providing matching communication skills. Patient input gathered during the needs assessment was used to operationalize the overarching objective in 11 intervention objectives that relate to the awareness, attitude, and skills of the patient (Figure 3, column 1). These objectives were linked to theoretical methods and corresponding practical strategies. The main criterion for the selection of the strategies was the ability to operationalize strategies in an online environment that could be hosted by Hematon. Three theory-based methods and four practical strategies were selected to influence the attitude and skills of patients (Figure 3, columns 2 and 3).

The main method chosen was modelling. Modelling has proven to be effective in patient-targeted skill building interventions [30-33] and can be operationalized in an online environment by means of video clips. Moreover, pre-visit goal setting was selected to encourage patient involvement during the consultations. This strategy was operationalized in two ways. First, the patient's appointment dates were linked to a reminder system, which reminds patients a week before their consultation to access the online intervention in order to prepare for their visit. Second, an open QPS was integrated, which could be completed and printed or sent to one's personal email address. A QPS can enhance the contribution of patients in medical communication [34-36]. Finally, there was an option to store, play back, and share audio recordings of a consultation with relatives, via their personal account. Playing back audio recordings has been shown to enhance recall, improve informed decision making, reduce anxiety, and improve communication with family members [37,38].

The intervention objectives were based on the experiences expressed by the patient service users. However, because this was a more theoretical phase comprising the literature search and the analysis of data, further patient involvement in this step was limited to a discussion with the patient research partners. The outcomes were presented and the feasibility of the operationalization was discussed, which was important for the final implementation. Their feedback did not change the initial outcomes.

Intervention Design (Step 4)

The goal of the fourth step was to design the content, structure, and layout of the intervention, inspired by the information gathered in the previous steps. An iterative design method was applied, that is, intermediate results (eg, video scripts, website navigation) were presented to patients and experts.

Subsequently, the intermediate results were adapted to their feedback, which is discussed in the following paragraphs.

The targeted intervention objectives (Step 2) were translated into five video diaries, in which five simulated patients demonstrate different communication skills. Each video diary (see Multimedia Appendix 2) displays the story of one lymphoma patient in 11 to 12 short clips (47-180 seconds). This setup was chosen to capture the experiences of a large group of patients and incorporate them in five personal stories, whereas a selection of only five patients may provide a biased view [39]. The scripts for the video clips were based on personal stories that patients had expressed in Step 1. Additional material was gathered with video recordings and real-time observations of relevant hematologic consultations. This type of patient contribution represents the lowest step in the participation ladder as the involved patients agreed to be observed, but had no further understanding of the project. Subsequently, a patient research partner reviewed the scripts. The feedback contained suggestions and corresponding content for additional scenes and unclear medical/technical jargon was highlighted. We incorporated the additional scenes in the video clips and rephrased the highlighted sentences. After recording the clips, the rough material was shown to a physician, an ICT expert, a patient research partner, and an external communication researcher. Their feedback was used in the editing process. For example, the reactions of the doctors to patients' communication behavior were cut out as a result of the feedback, aiming to increase the focus on the modelled communication behavior of the patient.

Given the changing preferences and needs of the patient, the working group chose not to present all 58 video clips to the patient at once. The patient-perceived, pre-visit communication needs determine the selection of three most relevant objectives, leading to the matching video clips. These needs are measured with an adapted version of the 10-item, 5-point Likert scaled Perceived Efficacy in Patient-Physician Interaction instrument (PEPPI). In this scale, patients indicate pre-visit their expected efficacy and post-visit their perceived momentary efficacy in communication [40]. Based on the input of patients, two extra tailored variables were added to determine which two video diaries match the patient's situation best: (1) the patient's preference to visit their HCP alone or with a companion, and (2) the stage of treatment (ie, ahead of treatment, in the middle of treatment, in remission, cured but monitored, and wait-and-see policy). If a patient wants to prepare his or her next consultation, new clips will be selected and these will be added to their previous selection.

Special attention was paid to two aspects that can influence the uptake of eHealth interventions: usability and credibility. According to Nielsen, usability is a quality attribute that assesses how easy user interfaces are to use and it is a necessary condition to bind users to a website. Credibility is an important element for the persuasive character of the intervention [41]. To enhance the persuasive character of the intervention, the Stanford Guidelines for Web Credibility were followed [42]. After testing preliminary versions of the intervention, a more comprehensive credibility and usability evaluation was performed by experts and prospective users. A heuristic evaluation (expert-based) and a think-aloud procedure (user-based) were set up with a total of 8 participants, which should be enough to detect over 80% of the usability problems [43]. A heuristic evaluation involves having a small set of evaluators examine the interface and judge its compliance with recognized usability and credibility principles (the heuristics). The list of heuristics used to evaluate PatientTIME was composed with the 10 usability criteria of Nielsen, supplemented with usability criteria specifically developed for older Web users [44], who are expected to be over-represented in the targeted population. The Stanford Guidelines for Web Credibility were added to this list, in order to objectively evaluate the aforementioned Web credibility. The list included themes such as consistency, user control, and efficiency. Three software experts and one master graduate in communication individually evaluated the intervention based on the list of heuristics. The user-based test included a think-aloud procedure. Two patients and two healthy people were asked to perform a set of consecutive tasks, which represented the major functionality of the intervention. Simultaneously, the subjects were encouraged to verbalize their thoughts [45]. Participants of both tests were asked to suggest improvements about the issues they came across.

The main credibility and usability issues that were identified are summarized in Table 1. Changes to these issues were incorporated before the release of PatientTIME apart from one. The illustrative pictures of patients in the layout were evaluated by the users as too positive. However, because we wanted to present a positive and encouraging context, we kept these pictures.

Table 1. Summary of identified credibility and usability issues.

Identified issues		Processed changes
Credibility	Information about collaborating parties, help function, and privacy issues is missing / unclear.	An extra information page was added with separate tabs containing information about collaborating stakeholders, introducing members of the working group, explaining privacy issues, and explaining the scientific context. A separate 'help' function was highlighted with contact details, frequently asked questions, and a project summary.
Functionality	Print function QPS unclear and use of the agenda not clear.	The agenda was made accessible on the home page, corresponding text was changed, and buttons were highlighted. The print function of the QPS[a]was highlighted.
Navigation	Location and additional text related to 'log-in' button is confusing. It is not always clear which elements are 'buttons'. Not always clear where you are in the website.	The consistency in color use and type of buttons improved, more contrasting colors were used when mouse-over, headings of active pages remain highlighted and stand out more comparing to the headings of inactive pages, the home pages present instructing messages to the user about the project status.
Information	Some texts are too formal. Some inconsistency in use of terms / jargon.	Textual changes were made.
Layout	Illustrative pictures too positive / happy. Unclear presentation of the selection of video clips. It is not clear what the content of the 'video archive' is or will be.	Another way to present the video diaries was developed, the video archive was removed and its function was incorporated in the video page.

[a]QPS: question prompt sheet

Development of Evaluation Plan (Step 5)

The goal of the fifth step was to develop an evaluation plan to examine the effects of the intervention. Decisions regarding the evaluation were partly stipulated in the research protocol, which proposed a randomized controlled trial (RCT) in which participants are randomized into the intervention group (with access to PatientTIME) and control group patients (without access to PatientTIME).

While working out the RCT protocol, practical issues like recruitment and patient information were discussed with the research partners and questionnaires were developed in collaboration with them. One patient research partner and one patient service user were asked to pre-test the developed questionnaires with a think-aloud procedure. Their feedback focused mainly on questions initially formulated as too formal or medical jargon that was unclear.

The involvement of prospective participants (ie, patient service users) in the RCT was planned on different levels. Both intervention and control group participants were asked to participate for a maximum of three consultations and they were both asked to fill in questionnaires delivered via their personal account. On the lowest participation level, participants are provided with information and asked to complete questionnaires. On a second level, they are encouraged to verbalize their ideas and input with regard to the study design to inform decisions taken by the working group. Last, a random subset of patients in the intervention group is encouraged to audio record and upload their consultation(s) on their secured PatientTIME account. This pilot was designed for the purpose of evaluating the playback option as well as to be analyzed by the researchers on their actual participation during their consultation.

The developed RCT protocol was audited with external experts to evaluate privacy issues and the exchange of online information and to assess and reduce possible risks. Because of the juridical, technical nature of the audit, we did not include patients in this audit. The Medical Ethical Committee of the Radboud University Nijmegen Medical Centre evaluated the RCT protocol and concluded that following the Dutch Medical Research Involving Human Subjects Act, the study did not require ethics approval. The RCT (registered in the Netherlands Trial Register, 3779) started in 2013 and the first results are expected to be available in 2015.

Development of Implementation Plan (Step 6)

The goal of the last step was to design an implementation plan that would guide the transfer of the intervention to a publicly available online tool. Contrary to the detailed evaluation plan, the implementation plan was a rough setup of actions that were guided by and adapted to decisions made in previous steps. To increase the chance of a successful implementation and adoption, the involvement of patients and Hematon in the planning and execution of the actual implementation started as early as the project planning. In the preparatory step, the board of Hematon was asked to help thinking about the valorization of the research results. In this way, we aimed to divide responsibilities at an early stage and awareness was created about the upcoming intervention.

Hematon wanted to make developed materials available for all their members and other patients. As a result, an agreement was established noting that after research is finalized, Hematon would become responsible for hosting the tool. Subsequently, during the development of the intervention and evaluation plan, several meetings were planned with our software developer and the webmasters of Hematon. In consultation with them, we aimed to develop materials that were not only usable for the secured trial setting, but could easily be transferred to a publicly available tool. Both patient research partners will be actively involved in the actual transfer of the intervention.

This transfer is not within the scope of this paper and will be done when the RCT proves to be acceptable, usable, and

efficient. Lessons learned from the evaluation will be used to optimize the intervention before implementation.

Discussion

Principal Findings

In the PatientTIME project, patients were given the opportunity to actively participate in the development of an online communication intervention with corresponding evaluation and implementation plan. In conformity with previously publications, the cooperation with patients brought valuable insights and appeared to influence many decisions made [46,47]. By combining patient participatory methods with a theoretical protocol, we aimed to create a bottom-up inspired development procedure. We encountered both facilitating elements, as well as obstacles in this approach.

Facilitators to Participatory Development

The combination of evidence-based and patient participatory methods did assist us in involving patients. The structure of the IM framework helped us choose when to involve patients, while the idea of participation ladders and user-centered design thinking inspired us in how to involve patients.

The involvement of patients on different levels appeared to be useful and practical. The patient research partners ensured a continuous patient-centered view, while the patient service users were able to give fresh new insights on different protocol steps.

Both Hematon as well as the research partners were involved from the very beginning of the project as a result of the preparatory planning step. We experienced this as a precondition to creating a continuous patient-centered view. Their early involvement supported the participation of patient service users and it gave the opportunity to discuss possible valorization of results at an early stage.

Another facilitating aspect was the attitude of the participating patients. They all seemed to recognize why the intervention was developed. This appeared to be a driving force behind their motivation to participate. Attracting engaged patients may be a precondition to creating a successful patient-centered approach.

Obstacles to Participatory Development

The recruitment and involvement of patients was a time-consuming part of the project. In some steps, we could have benefited from more involved patient service users (especially the intervention development step), but time constraints prevented us from doing so. The extent of patient involvement relates to the amount of time available to execute the project. However, we think time constraints should not be a reason for limited participation.

Flexibility in terms of planning and setup seemed a precondition to including the perspectives of the (seriously ill) patients. For example, during the needs assessment, some patients were too ill to attend a focus group session. An interview at their home gave us the opportunity to incorporate their experiences as well. Considering the illness of the targeted patients, we think the extent of involvement of service users should be evaluated per protocol step.

Flexibility also appeared to be a key concept in incorporating patients' viewpoints and experiences in the defined research proposal. In the current study, a research proposal defined certain decisions, for example, the intervention would be delivered online and the evaluation of the effects would be tested in an RCT. Although the proposal was built on previous research and experiences, these decisions were made before the targeted patients could be consulted (see Future Research).

Future Research

While there is a desire for more patient participation in research, it seems to clash with strict research proposals and protocols that need to be approved before the start of a project. Perhaps researchers should involve (ex-) patients in the design of such documents. However, this still does not give the required flexibility to adapt a project to the input of patients, gathered along the way. Patient participation in research projects that include design activities requires methodologies that allow the dynamics of design (eg, by patient input) to influence the process. Intervention mapping can be a guiding method, unless it is bounded to a strict predefined proposal. Participatory Learning and Action Research or Design Inclusive Research might be interesting alternative methodologies [48-50]. Funders also should evaluate the extent of detail they request in proposed projects and how this might restrict the extent of (true) influence patients can have.

Considering the evaluation of online interventions and the necessary flexibility to incorporate patients' input, it might be interesting to study other perhaps more flexible evaluation methods than an RCT. A longitudinal study where intermediate results can be used to optimize the intervention during the test phase might be an interesting alternative. Furthermore, some patients might have a strong preference for using or not using technology. In the case of strong preferences, results may be biased when using a regular randomized controlled trial. Within preference trial designs, this bias is dealt with by the fact that patients with strong preferences for either intervention will get the intervention they prefer. Only those without explicit preference are randomly assigned to either the intervention or the control group [51].

Limitations

A limitation of the applied method is that the participating patients represent a self-selected convenience sample as involved patients voluntary signed up to contribute to the study. This could have led to a biased view of a more empowered group of patients. In general, the possibility of having a biased group of participants in a participatory development approach is evident, as one needs to find patients that are interested in cooperating. On the other hand, one wants to develop an intervention that reaches out to the whole targeted population. This advocates the use of different participation levels and creative solutions to attract and/or select patient service users to capture a broad view of experiences.

Similar to other studies [52], in the current study the IM framework was not applied in a linear way as proposed, which

can be argued as a potential limitation. However, a design process rarely follows a parallel execution process and, especially because the aforementioned flexibility was required, we think it does not have to affect the quality of the developed products.

Conclusions

Involvement of patient research partners in combination with patient service users can inspire and guide the evidence-based intervention mapping protocol. Early involvement, involvement on different levels, and flexibility in terms of planning and setup seem to be preconditions to create a bottom-up inspired development procedure with (seriously ill) patients. Further research is necessary to find out if a more patient-centered approach improves the implementation and uptake of eHealth interventions.

Acknowledgments

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Conflicts of Interest

None declared.

Multimedia Appendix 1

Intervention.

[PDF File (Adobe PDF File), 83KB - resprot_v3i4e59_app1.pdf]

Multimedia Appendix 2

Video diaries.

[PDF File (Adobe PDF File), 75KB - resprot_v3i4e59_app2.pdf]

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Abbreviations

HCP: health care professional
ICT: information and communication technology
IM: intervention mapping framework
PatientTIME: Patients Talk in Medical Encounters
PEPPI: Perceived Efficacy in Patient-Physician Interaction
QPS: question prompt sheet
RCT: randomized controlled trial
UCD: user-centered design

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Review

“Real-World” Practical Evaluation Strategies: A Review of Telehealth Evaluation

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Abstract

Background: Currently, the increasing interest in telehealth and significant technological breakthroughs of the past decade create favorable conditions for the widespread adoption of telehealth services. Therefore, expectations are high that telehealth can help alleviate prevailing challenges in health care delivery. However, in order to translate current research to policy and facilitate adoption by patients and health care providers, there is need for compelling evidence of the effectiveness of telehealth interventions. Such evidence is gathered from rigorously designed research studies, which may not always be practical in many real-world settings.

Objective: Our aim was to summarize current telehealth evaluation strategies and challenges and to outline practical approaches to conduct evaluation in real-world settings using one of our previously reported telehealth initiatives, the Diabetes Connect program, as a case study.

Methods: We reviewed commonly used current evaluation frameworks and strategies, as well as best practices based on successful evaluative efforts to date to address commonly encountered challenges in telehealth evaluation. These challenges in telehealth evaluation and commonly used frameworks are described relevant to the evaluation of Diabetes Connect, a 12-month Web-based blood glucose monitoring program.

Results: Designers of telehealth evaluation frameworks must give careful consideration to the elements of planning, implementation, and impact assessment of interventions. Evaluating performance at each of these phases is critical to the overall success of an intervention. Although impact assessment occurs at the end of a program, our review shows that it should begin at the point of problem definition. Critical to the success of an evaluative strategy is early planning that involves all stakeholders to identify the overall goals of the program and key measures of success at each phase of the program life cycle. This strategy should enable selection of an appropriate evaluation strategy and measures to aid in the ongoing development and implementation of telehealth and provide better evidence of program impact.

Conclusions: We recommend a pragmatic, multi-method, multi-phase approach to telehealth evaluation that is flexible and can be adapted to the characteristics and challenges unique to each telehealth program.

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KEYWORDS

telehealth; eHealth; evaluation; evaluation framework; diabetes mellitus; technology

Introduction

Background

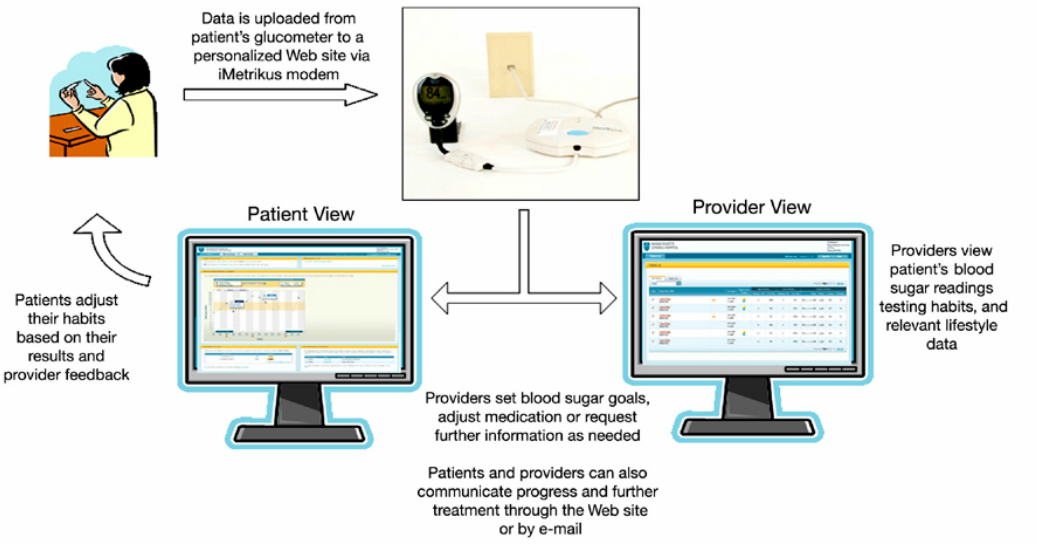
Current global trends suggest that the popularity of telehealth is at an all-time high since the advent of modern telehealth over 40 years ago [1]. This rise is attributable to expectations that telehealth can help improve the current health care conundrum—rising prevalence of chronic diseases, shortage in health care workforce, and rising health care costs. These demands coincide with tremendous advancements in the technological landscape. Today, due to increasing affordability and user-friendliness of modern technologies, they have become ubiquitous. For example, more than 85% of the world's population today has a mobile phone [2]. This abundance makes mobile phones a ready medium for health care delivery, capable of uniquely engaging patients to improve quality and access to care. Telehealth interventions come in various forms, ranging from simple one-way text messaging aimed at health education, to the collection and transmission of relevant biometric data used in home monitoring programs [3-5]. More recently, systems driven by complex algorithms using sensors and evidence-based psychological theories to motivate sustained behavior change are being developed, and early tests find evidence of better clinical and economic success [6].

Over the last decade, support has been growing for the use of telehealth. The ongoing health care reforms have significantly increased government funding as well as increased state legislation supporting expansion of telehealth services [7,8]. Consequently, adoption has also increased among insurers, health care providers, and professional associations. For example, home monitoring of blood pressure and blood glucose (BG) are now basic components of hypertension and diabetes management guidelines [9,10].

Despite the great strides made in the field, critics argue that evidence of the impact of telehealth is not strong [11]. The evidence of an intervention's impact has been traditionally established by systematically and rigorously designed research studies. Randomized experiments that control for a variety of biases remain the gold standard for such evaluations. Policy makers often base their decisions on research from a meta-analysis of multiple evaluation efforts, where individual study estimates are pooled to estimate the overall effect of similar telehealth intervention programs. However, current telehealth literature is largely heterogeneous consisting of many pilots and mixed quality trials with diverse outcomes, which makes it difficult to estimate pooled effects of telehealth [12]. The time and resources required to test telehealth programs in randomized experiments is substantial, and with rapid changes in technology, impractical. In this paper, our goal was not to propose a new evaluation framework but rather to summarize current evaluation strategies in telehealth and outline practical steps to conducting evaluation in real-world settings using one of our previously reported telehealth initiatives, the Diabetes Connect program (DC), as a case study.

Diabetes Connect is a 12-month Web-based blood glucose monitoring program. The program is designed to help improve blood glucose control by promoting patient self-management in the area of regular self-monitoring (a recommended diabetes self-care behavior) and facilitating patient-care provider connection (Figure 1). The program enables patients to easily collect and upload their blood glucose readings, via data transfer devices, to a secure Web-based platform. The real-time data sharing enables the care team to provide personalized and timely feedback to patients. Findings from this program are reported in previous publications [4,13,14]. The program was evaluated through all the stages of a telehealth project life cycle, and we found it helpful to follow the simple evaluative life cycle model described subsequently. Before describing the methods and results of the DC evaluation, we will briefly summarize some commonly used current evaluation frameworks and strategies.

Figure 1. Diabetes Connect conceptual model.



Overview of Current Evaluation Work, Challenges, and Emerging Frameworks

A number of program evaluation frameworks are available, depending on the phase of program development and the feasibility of data collection. In this paper, we provide a brief summary of challenges in telehealth evaluation and commonly used frameworks relevant to the evaluation of DC to orient investigators new to telehealth project design and program evaluation. For a comprehensive review of existing telehealth evaluation frameworks, see Ekeland et al [12] and van Gemert-Pijnen et al [15]. Bashshur et al describe four types: (1) evaluability assessment, (2) documentation evaluation, (3) formative or process evaluation, and (4) summative or outcome evaluation [16]. Each evaluation type is defined in Table 1 with an example based on DC.

Table 1. Evaluation typology.

Type	Definition	Key features and uses	Example
Evaluability	Assessment conducted prior to or at the beginning of a program to make explicit the goals and objectives of the program and intended effects or outcomes	Frame research question Determine research design Identify measurement tools and data collection methods Determine analytic methods	Stakeholders (eg, investigators, health care professionals, case workers, and patients) meet to discuss goals of the program and identify key processes and outcomes that the program is intended to impact.
Documentation	A narrative description of the implementation of the program	Description of procedures and protocol used Description of difficulties encountered Description of steps taken to address barriers to implementation Identify successful strategies to dealing with barriers Enable others to reproduce the program in other settings	The program is to be used at four clinics. Notes are taken on how the program is implemented across sites, barriers or difficulties to implementation, and any modifications to the overall program or site-specific adjustments.
Formative or process	Evaluation focusing on the effects of the program on the process of care	Behavioral and attitudinal changes related to program adoption and use Identify barriers to adoption and use Resolve workflow integration issues Identify technical problems	A formative evaluation using interviews and focus groups with patients was conducted to examine if DC improved disease management, communication between patients and providers, and whether there were any unexpected barriers to effective use.
Summative or outcome	Provides evidence of the intended effects of the program.	Robust evidence of program effects Identify benefits of a program Provide evidence to decision makers and policy makers of a program's benefits	The key objective of the DC program was to augment care with home monitoring of blood glucose. Summative evaluation was conducted to examine the effect of the intervention on clinical outcomes measured by HbA1c.

To date, most telehealth research has been small-scale studies, and evaluation research has largely been formative and focused on process variables [16]. Although formative evaluations do not provide generalizable results on the impact of telehealth, they do provide useful information on the behavioral, attitudinal, and cognitive factors related to program implementation and subsequent outcomes [16].

There have been calls for larger, long-term telehealth studies that would provide measures amenable to summative evaluation examining the intended effect of a program. Perhaps the most important outcomes are health care costs, and the potential benefits and gains in efficiency that telehealth might bring to health care delivery processes [16]. Summative evaluations most often employ randomized controlled trial (RCT) designs and standardized measures to generate results that can be compared across studies.

There are many issues that complicate the ability to conduct telehealth program evaluations, particularly robust summative evaluations. We summarize these issues into three interrelated challenges: (1) the diversity of telehealth programs, (2) traditional research designs like RCTs are often impractical for telehealth evaluation, and (3) telehealth programs are complex and dynamic, and evaluation frameworks do not capture all process and outcome metrics satisfactorily.

The first challenge is that telehealth interventions are diverse and span a range of medical conditions, health care delivery

problems, and types of intervention strategies, thereby making it difficult to specify the parameters of a standard telehealth program. This problem sometimes makes outcome metrics program specific and restricts the generalizability of findings. This is further complicated by the insufficient description of many telehealth interventions, which makes reproducibility and comparison of interventions very difficult [17]. The CONSORT-EHEALTH elaborates on this challenge and recommends a checklist to standardize reporting of component parts of telehealth interventions [17]. Also, technology is constantly changing and creates new opportunities for innovative programs. As a result, the field of telehealth is in "constant flux" [16] and is difficult to define. In addition, as telehealth is integrated more broadly as part of regular care delivery, it becomes more difficult to define telehealth as a distinct modality of care [16].

A second challenge is that an RCT, the most widely accepted evaluation methodology, is often impractical for telehealth programs [16,18]. The classic experimental RCT design features the randomization of subjects to intervention or control groups, carefully concealing or "blinding" subjects and investigators to intervention assignment, and pre- and post-intervention measurements. Evidence from RCTs provides the most robust method to establish cause and effect relationships and is less likely to be influenced by subjects or investigators. However, a number of issues limit the use of RCTs to evaluate telehealth. In many cases, the assignment to control or intervention cannot be concealed from subjects, health care professionals, or investigators. RCTs are also expensive and labor intensive to conduct, which often limits studies to small samples and short study durations [19]. Additionally, the rapid pace of changes in technology means that programs are sometimes obsolete by the time a long-term RCT has been completed. Other problems with how telehealth is implemented can introduce heterogeneity in the intervention and observed outcomes. For example, workflow and staffing across multiple sites is difficult to control and differences between sites can influence the observed effects [16].

The third challenge to telehealth evaluation is that health programs are more complex and dynamic than other types of interventions [16,20]. The evaluation of medication efficacy, for example, is a more clearly defined cause and effect relationship that is amenable to an experimental design. The success of complex health interventions like telehealth, however, is dependent on human behavior, program adoption, level of participant activation or engagement, and other contextual factors present where the intervention is implemented [20].

In response to these challenges, many current frameworks conceptualize telehealth as a complex health intervention that undergoes several stages of development, testing, and deployment. In addition, these frameworks highlight the role of stakeholders (eg, patients, caregivers, health care professionals and administrators, insurers, and state and federal agencies) and contextual factors in determining success. Khoja et al propose a framework that spans a telehealth program "life cycle" consisting of four phases: (1) development, (2) implementation, (3) integration, and (4) sustained operation [21]. Across the program life cycle, they identify several evaluation themes and associated outcomes. For example, during the implementation phase, one evaluation theme is "health services" with outcomes that include (1) improved diagnosis and treatment, (2) improved decision support, (3) better clinical safety, and (4) equity of care. Van Gemert-Pijnen et al propose a "holistic" framework that highlights the role of stakeholders as active participants in the development, implementation, and evaluation of telehealth programs [15]. The first step is a contextual inquiry to gather information from stakeholders to identify needs and gain insight for finding solutions. Findings from this step are further elaborated on by a value specification to identify the most favorable solutions. Through a process of continuous, iterative formative evaluations, a telehealth program can be tailored to fit the needs of users and the health care context. As the program matures, a summative evaluation is used to measure the success of the program based on the key goals and measures identified at the beginning of the project.

The RE-AIM Evaluation Framework (see Table 2) has been used to evaluate telehealth as well as other health programs [22-24]. It focuses on both the individual (Reach and Effectiveness) and organization level (Adoption, Implementation, and Maintenance) metrics to assess program impact. It translates research findings into action by encouraging stakeholders to focus on essential program elements and their external validity, not just the outcome of the research like many traditional models [22]. This enhances the quality, speed, and impact of the research in the real world, in turn, creating effective, generalizable, and evidenced-based interventions. Although the RE-AIM framework lacks a clear progression of program phases, it provides a flexible framework that identifies five elements common to health programs that determine success.

Table 2. RE-AIM elements and definitions.

RE-AIM element	Definition
Reach	The number and percent of people from the target population who participate, and their representativeness.
Effectiveness	The change in outcomes observed over the duration of the intervention.
Adoption	The number and percent of settings and staff who are expected to use the intervention and who participate.
Implementation	The extent to which the intervention is delivered consistently and the time and costs associated with implementation.
Maintenance	The long-term effects on key outcomes, and the extent to which a program is sustained, modified, or discontinued after the initial trial phase.

Rather than creating new methods for telehealth evaluation, many authors recommend greater standardization of existing methods and the use of multiple or mixed methods. Where possible, the standardization of study design, outcome measures, and analytic techniques would improve the ability to compare results between telehealth programs and conduct meta-analyses aimed at evaluating the field as a whole [12]. However, standardization of methods across the many types of telehealth programs has been difficult to achieve. To generate more robust conclusions of causality, the use of multiple methods, using both qualitative and quantitative research methods [25], and frameworks to "triangulate" the effects of telehealth programs have been suggested as a useful strategy [16].

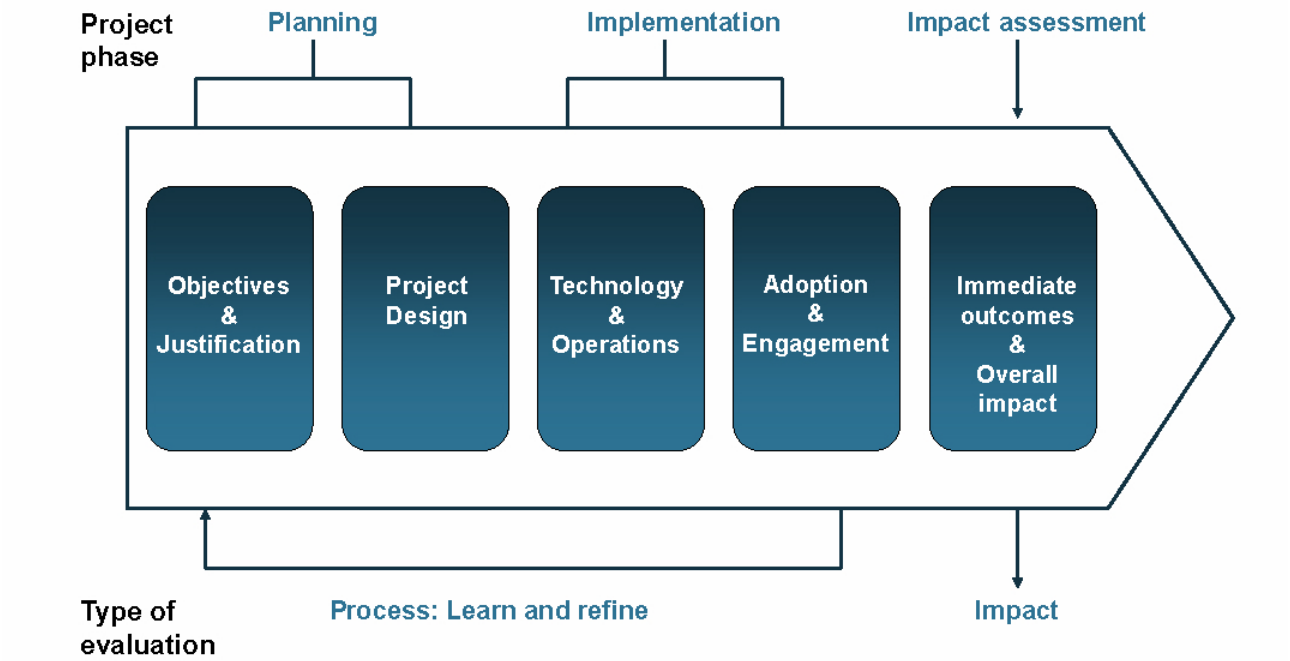
In sum, these challenges highlight the need to carefully consider a practical set of goals and strategies for conducting telehealth evaluations in "real-world" settings, given the limitations of available resources and opportunities to collect data.

Methods

A Practical Approach to Conducting "Real-World" Telehealth Evaluation

We describe some best practices based on successful evaluative efforts to date to address commonly encountered challenges in telehealth evaluation: time, impracticality of RCTs, and the heterogeneity of implementation and diversity of outcomes across the program pathway. In its simplest form, the life cycle of any project involves three main phases with several activities: planning, implementation, and impact assessment (Figure 2). Evaluating performance at each of these phases is critical to the overall success of an intervention. Although impact assessment, the overall measure of effectiveness of the intervention, occurs at the end of a program, it should not be an afterthought but should begin at the point of problem definition. For this reason, fidelity to project protocol in implementing all three phases is a necessary prerequisite for a valid and methodologically sound evaluation [26].

Figure 2. Telehealth program pathway.



Planning

Objectives and Justification

The first step in planning is to define the goals and set the direction for the project using guidelines for SMART (Specific, Measurable, Achievable, Relevant, and Time-bound) goals and objectives [27]. Goals may be based on improving outcomes, increasing access, decreasing costs, or may simply be learning objectives and should guide the generation of performance indicators to evaluate the project. They are usually based on what may be of interest to funders and stakeholders, especially those who will accelerate the deployment of these programs at scale. At this stage, a consultation with a biostatistician may be necessary in specifying measurable endpoints. Our objectives were to determine the effect of the program on clinical outcomes (measured by changes in HbA1c, trends in BG measurements). Being one of our first experiences with remote monitoring for diabetes, we were also interested in general usability, satisfaction, and determinants of participant engagement in the program.

Project Design

Based on the outcomes expected, processes and activities required for project implementation should be defined. This includes identification of the target population, identification of stakeholders and role definition, design of project protocol, budgeting, staff allocations, setting timelines, etc. One must keep in mind that performance indicators should be generated for each step in the process.

After relevant outcome measures are agreed upon, a suitable evaluation design should be identified. Some goals, like engagement or adoption metrics, can be interpreted without any

comparison data. However, for most other goals, including access and clinical outcomes, a comparison arm is required. Randomized control groups are considered to be the gold standard, but a variety of other methodologies have been used in telehealth evaluation, albeit with certain limitations. Paired analyses before and after the intervention, matched control analysis, as well as using existing population-level metrics for comparison have all been used with varying success [28]. These options should be considered when faced with time or monetary constraints in using an RCT approach.

Implementation

This is usually the longest phase in a project and consists of assigning inputs and generating outputs for project execution.

Technology and Operations

Project objectives and design will inform the decisions that need to be made about technology and operational processes, which are the main inputs needed to perform project activities. Inputs, by definition, are the resources needed to support the primary activities of the project, and they include funding, staff and their expertise, technology, and other materials needed for project implementation. During this time, careful consideration must be given to select the "right" technology to develop an effective telehealth intervention [18] and also to ensure that all operational processes conform to the pre-specified plan. All protocol deviations should be documented and reported to the principal investigator and appropriate institutional review board staff in a timely fashion. Furthermore, the technology should be evaluated for its ability to function as specified and integrate adequately with other technologies and existing systems, as well as it timely availability, usability, and acceptability. In addition, it is also helpful to be vigilant to disruptions in workflow processes or system malfunction. These and other technical problems reported by users should be logged, reported, and addressed promptly.

Once the technology has been chosen, deployment strategies can be designed. Program adoption and engagement can vary widely based on who offers the technology to patients, how it is set up, and the availability of customer service after initial deployment. Measures for each of these assumptions and requirements should be made part of the evaluation plan.

Adoption and Engagement

Strategies relating to major stakeholder (patient and provider) engagement and adoption are critical elements of a telehealth intervention. Rogers describes an "innovation-decision" process involved in adopting or rejecting a new intervention [29] and defines adoption as the decision of stakeholders to make full use of an innovation as the best course of action available [29]. Subsequently, stakeholders begin to engage with the intervention. Engagement refers to "actions individuals must take to obtain the greatest benefit from the health care services available to them" [30]. Evidence exists that provider and patient engagement are interlinked, and thought must be given to measuring both appropriately. For patients, themes to include are literacy, intrusiveness, user interface design, simplicity, and usability of intervention. For providers, integration with existing workflow, appropriateness of personnel delegation, electronic medical record integration, and ease of program deployment should be considered. Also, behavioral themes like activation and readiness to change are critical factors in optimizing engagement. Examples of techniques to improve user engagement include personalization, behavioral economics (like rewards/incentives, nudges), gamification, and social networks.

Impact Assessment

This phase includes measurement of both intermediate outcomes and overall effects (impact assessment) of the intervention. Intermediate outcomes are direct effects attributable to project outputs, like increased knowledge, increased adherence to recommended treatments, behavior change, early detection, and treatment of symptoms. Often they may be used as proxies to measure endpoints. In cases where objective measurements of effects may be difficult to assess quantitatively, common qualitative approaches, like patient interviews or focus groups, can be used to more fully measure the effects attributable to the intervention and are particularly useful for assessing subjective patient preferences.

Impact assessment, on the other hand, estimates the net impact and overall effect of the intervention. They may be clinical or financial and are usually more meaningful to stakeholders. As a rule of thumb, the more rigorous the research methodology, the more accurate the effect estimate. However, in practice, the most rigorous design is not usually feasible. The goal should be to achieve a balance between managing real-world constraints and the next best possible design. Therefore, based on the setting and other limiting factors where the telehealth intervention is deployed, alternative study designs, which maximize efficiency, validity, and reproducibility while minimizing risk of bias should be considered (Textbox 1).

Textbox 1. Strategies to minimize risk of bias.

- considering multi-site trials to address the problem of small sample sizes [18]
- using validated questionnaires/tools to assess impact [18]
- using unobtrusive data collection techniques or routine clinical data [18]
- using both quantitative and qualitative approaches to completely and accurately estimate effects of intervention [12]
- considering formative approaches to improve next iteration of the project to increase scalability potential and maintain ongoing success
- using alternative research designs including quasi-experimental designs, matched control trials, staged interventional trials, crossover trials, etc [28]

Results

Planning: Objectives and Justification

In our DC program, we were interested in whether augmenting regular care with home monitoring of BG (thereby enabling patient self-management and providing clinicians with home BG data in between office visits) improved clinical outcomes as measured by change in HbA1c. This goal definition further helped us to specify program objectives and outcome variables. Our objectives were to determine the effect of the program on clinical outcomes (measured by changes in HbA1c, trends in BG measurements). Being one of our first experiences with remote monitoring for diabetes, we were also interested in general usability, satisfaction, and determinants of participant engagement in the program.

Implementation

Technology and Operations

In DC, the adopted technologies integrated well with existing systems and data transfer was confirmed within 24 hours of set-up. Technical personnel were available during business hours to answer questions or help troubleshoot for ongoing problems. Tracking the frequency of troubleshooting calls was particularly important for us to identify some differences early in the study in the pattern of participants' interactions with the technologies. We also found it very important for staff to standardize all practices, time log all problems, and track them until resolution. These processes were important for continuous system improvement.

A trial of the initial version of the DC in a pilot phase before mass enrollment of participants helped us identify early any problem areas in deployment, adoption, and engagement that required further work.

Adoption and Engagement

In this phase, some of the key evaluation questions in DC included:

- Was the target audience appropriately reached?
- Has the program been adopted by key stakeholders?
- Are key stakeholders engaged?
- Are engagement patterns different based on time, demographic group, or location?
- Are some participants responding differently to engagement techniques than others?
- Are ongoing troubleshooting issues reported and addressed promptly?

DC was adopted by clinicians in four different practices within the Partners HealthCare network of hospitals who enrolled suitable patients in the program for 12 months. Help desk support was available (Monday to Friday, business hours) to address in a timely fashion any technical issues that might arise. Patient engagement was assessed by number of uploads and BG readings recorded by the DC. We also assessed provider and practice engagement by assessing the number of times providers logged on to the Web portal to view patients' BG readings and the average number of provider logins to the Web portal by practice.

Impact Assessment

We used focused groups as a measure with DC because they allowed us to capture contextual content on participants' experiences, understandings, perspectives, or stories we might have otherwise missed in personal interviews. They facilitated lively conversations around usage, interactions with clinicians, and difficulties they encountered as they interacted with the system. We found focus groups particularly helpful to evaluate program perception, engagement, and adoption. From these groups, we discovered that most participants' favorite part of the program was the ability to easily send BG readings to their providers and when appropriate, they appreciated the timely feedback. Additionally, participants reported that their provider's interest in the program was the strongest predictor of whether or not they would use the program. These findings would have been nearly impossible to discover without the focus group and were very helpful in future program development.

A critical assumption in impact assessment is the availability of high-quality data. We ensured that our endpoint data were collected appropriately and in a timely manner. Data points from DC include HbA1c, BG readings, frequency of data uploads, and frequency of logins to the portal. These data were vital for continued program improvement and outcome evaluation. For example, we found that the mean changes in HbA1c showed promising results: those who uploaded more BG readings had a further decline in their HbA1c. Additionally, the nature of DC made measuring engagement quantitatively through more than one metric simple, and we successfully measured the frequency of uploads by participants as well as logins by both participants and providers. We also used qualitative methods to assess our outcomes in participants recruited from four different sites. Through the focus groups, users and potential users of the program were interviewed to obtain feedback including comfort level using the system and how the program could be better tailored to their needs. Knowing what information was most important to collect during the program-planning phase made the impact assessment process easier to conduct, and we were able to detect meaningful changes in our outcome measures.

Discussion

Principal Considerations

We have provided a brief review of evaluation strategies, drawing on our experiences in creating and evaluating telehealth programs to describe a practical approach to conducting evaluations in real-world settings. We recommend that each telehealth program have an evaluation procedure in place at every stage of the program life cycle, from ideation to large-scale deployment and impact assessment, while considering the different characteristics and challenges unique to each telehealth program. Key to implementing this strategy is early planning involving all stakeholders to define overall goals and goals for each project phase. Once consensus is reached, an evaluation design should be selected that best fits the identified goals and the constraints of project resources.

Early planning is essential to ensure valid measurement of goals across program phases and to evaluate overall impact.

The telehealth evaluation strategy we described has two distinct advantages over classic, large-scale RCT studies. First, telehealth interventions are complex health interventions that can be best understood as "works in progress" tailored to fit the unique needs of each site and set of users. Telehealth evaluation across the program life cycle can provide useful data to guide program design and improve the reach, effectiveness, and adoption of new interventions. This evaluation strategy may accelerate the translation of research into practice to create programs that are well integrated into existing clinical workflows to improve efficiency and ultimately control health care costs. The results from formative/process evaluations are often key to developing telehealth programs with high levels of sustained user adoption and engagement. Patient engagement is an important objective in creating telehealth programs that focus on lifestyle change, improved patient self-management, and the prevention and control of chronic disease.

Second, greater participation by stakeholders early in the program planning process may foster sustained involvement and commitment to telehealth programs during implementation and large-scale deployment. This could increase the number of telehealth programs that are successfully transitioned to larger programs integrated into standards of care practices.

Some of the lessons we learned from DC and our other telehealth programs that have proven to be useful over the years are found in Textbox 2.

Textbox 2. Keypoints from Diabetes Connect and other telehealth programs.

- Start thinking about evaluation early and involve as many stakeholders as possible, including participants, funders, etc.
- Design your project based on proposed outcomes. Keep in mind the requirements to collect adequate data at the appropriate times and the need for objective metrics to evaluate each process in the program.
- Consider the limitations of the evaluation design. An RCT may not always be feasible, so explore other program design options, keeping in mind resource considerations and internal validity.
- Technology is essential, but not the focus of the program. Evaluate whether you are using the right technology based on ease and appropriateness of use, integration capabilities, and scalability. Remember that the final goal is to use something that your target population will adopt easily and engage with in the long term.
- Engagement in the program is key. Remember to include proven elements in your program design (like personalization, behavioral economics, etc) as well as metrics to evaluate their effectiveness in your evaluation plan.
- To maximize the potential of a valid, reliable evaluation, choose program design elements that reduce the potential for bias, like using validated survey instruments, using both qualitative and quantitative assessment methodologies, etc.

Conclusions

Efforts to evaluate telehealth programs create what Bashshur et al describe as a "quandary" [16]. On one hand, RCTs are the consensus method for conducting rigorous evaluation research. However, they are always not well suited or practical for telehealth evaluation [16,25,31]. We recommend a pragmatic, multi-method, multi-phase approach that is flexible and can be adapted to the characteristics and challenges unique to each telehealth program as showcased in the Diabetes Connect program. Critical to the success of this strategy is early planning that involves all stakeholders to identify the overall goals of the program and key measures of success at each phase of the program life cycle. Following this strategy should enable investigators to select an appropriate evaluation strategy and measures that will aid in the ongoing development and implementation of telehealth and provide better evidence of program impact.

Conflicts of Interest

None declared.

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Abbreviations

BG: blood glucose
DC: Diabetes Connect
RCT: randomized controlled trial

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Protocol

Gestational Age Assessment in the Ghana Randomized Air Pollution and Health Study (GRAPHS): Ultrasound Capacity Building, Fetal Biometry Protocol Development, and Ongoing Quality Control

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Abstract

Background: Four million premature deaths occur yearly as a result of smoke from cooking fires. The Ghana Randomized Air Pollution and Health Study (GRAPHS) is underway in the Kintampo North municipality and South district of rural Ghana to evaluate the impact of improved cook stoves introduced during pregnancy on birth weight and childhood pneumonia. These hypotheses are being tested in a cluster-randomized intervention trial among 1415 maternal-infant pairs within 35 communities assigned to a control arm (traditional cooking) or one of two intervention arms (cooking with an improved biomass stove; cooking with liquefied petroleum gas stoves).

Objective: The trial is designed to ensure delivery of the stove intervention prior to the period of maximal fetal growth. To answer questions about the impact of household air pollution on pregnancy outcome, accurate gestational age assessment is critical. This manuscript describes in detail the development of the gestational dating protocol, intensive ultrasound training involved, ultrasound capacity building, and ultrasound quality control program.

Methods: Ultrasound training occurred in several phases over the course of 2 years. Training included a basic obstetric ultrasound course offered to all midwives performing antenatal care at the two study hospitals, followed by a more intense period of hands-on training focused on fetal biometry for a select group of providers demonstrating aptitude in the basic course. A standard operating procedure was developed describing how to obtain all fetal biometric measurements. Consensus was obtained on how biometric images are used in the trial to establish gestational age and estimate the delivery date. An ongoing ultrasound quality control program including the use of an image scorecard was also designed.

Results: Publication of trial results is anticipated in late 2016.

Conclusions: Use of ultrasound should be strongly considered in field-based trials involving pregnant women to accurately establish gestational age, as menstrual dates may be incorrect or unknown. The inclusion of ultrasound in areas where ultrasound capacity does not previously exist requires a significant investment of time and resources. Such investment ensures appropriate training, high quality images, and accurate dating pregnancies. We outline our ultrasound training, image acquisition, quality control, and dating protocols in detail.

Trial Registration: Clinicaltrials.gov NCT01335490; http://clinicaltrials.gov/ct2/show/NCT01335490 (Archived by WebCite at http://www.webcitation.org/6UbERJNO6).

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KEYWORDS

ultrasound; capacity building; gestational age; biometry; household air pollution

Introduction

Background: The Ghana Randomized Air Pollution and Health Study (GRAPHS)

The majority of households in sub-Saharan Africa rely on solid biomass fuels such as charcoal, wood, or crop residues for cooking. Women, as the primary cooks, and their young children playing nearby are particularly vulnerable to harmful pollutants released from the inefficient combustion of these fuels. The resulting household air pollution causes nearly 4 million premature deaths per year and represents one of the major global environmental risk factors for reduced life expectancy, due in large part to an increased risk of death from acute lower respiratory infections (pneumonia) in childhood [1-3]. The overall objective of the Ghana Randomized Air Pollution and Health Study (GRAPHS; Trial Registration NCT01335490) is to test the effectiveness of improved cook stoves introduced prior to the third trimester in pregnancy to increase birth weight and reduce pneumonia in the first year of life (Jack et al, forthcoming). If effective, distribution of cleaner cooking technology through existing antenatal programs may be considered by policymakers to reduce household air pollution attributable deaths in early childhood.

Our hypotheses are being tested in a cluster-randomized intervention trial among 1415 maternal-infant pairs in the Kintampo North municipality and South district of rural Ghana in West Africa. In the 35 trial communities, pregnant women are being identified via community-based fieldworkers, taking advantage of an existing health and demographic surveillance system. Women confirmed by ultrasound to be carrying a single live fetus less than 24 weeks estimated gestation who consent to participation are enrolled into the trial. Enrolled women are followed during pregnancy and their children monitored through one year of life to assess the impact of the intervention on two primary endpoints (birth weight and acute lower respiratory infections). Within intervention communities, improved cook stoves are distributed to enrolled pregnant women prior to 28 weeks, along with health insurance and an insecticide-treated bed net. Women in one of the intervention arms will receive a two-burner liquefied petroleum gas (LPG) stove along with LPG, which will be supplied to them throughout the study duration. In the other arm, enrolled women receive two improved biomass stoves (BioLite, Brooklyn, New York). In laboratory settings, LPG and the BioLite stoves achieve reductions of 80% or more in combustion products. The impact in field settings from exposure reduction on health outcomes using these stove interventions has not been established.

Rationale for Accurate Gestational Age Assessment in GRAPHS

To ensure that our stove interventions are delivered prior to the third trimester when fetal growth is at a maximum, accurate gestational age assessment of the screened pregnant women is required. Furthermore, if we identify a difference in mean birth weight by intervention arm, we aim to determine whether this stems from a difference in infants born too early, too small, or both. These secondary trial outcomes (preterm birth and small for gestational age) also require knowledge of the gestational age. In the first half of pregnancy, ultrasound evaluation of gestational age is more accurate than menstrual dating [4-5]. Postnatal estimates of gestational age, and examinations such as the Ballard [6] or Dubowitz [7], are unable to verify inclusion criteria and may misclassify preterm infants as term or small for gestational infants as appropriately grown [8]. Menstrual recall is imperfect in our population. We therefore chose to incorporate ultrasound into GRAPHS at the time of enrollment using midwives at the study hospital. The feasibility of training mid-level providers to perform fetal biometry in resource-limited settings has been previously demonstrated [8-10]. In this manuscript, details of the ultrasound training program, the protocol for gestational age assessment in GRAPHS, and the ongoing quality review process are described. This will clarify how gestational age is determined for the trial and may be of utility to others embarking on similar efforts.

Methods

Ethics Approval and Funding

Ethical approvals for GRAPHS were obtained from the Institutional Review Boards of Columbia University Medical Center and the Massachusetts General Hospital/Partners Healthcare, the Ghana Health Service Ethical Review Committee, and the Kintampo Health Research Centre Institutional Ethics Committee. Funding for the trial is provided through the United States National Institute of Environmental Health Sciences (NIH 1R01ES019547) and the Ghana Ministry of Health.

Study Area

GRAPHS is taking place in 35 randomly selected communities in the Kintampo North Municipality and South Districts, which are located in the Brong Ahafo Region of Ghana. The two districts are largely rural and are monitored by an extensive health and demographic surveillance system [11]. Neonatal mortality is 32 deaths per 1000 live births and infant mortality estimated at 52 deaths per 1000 live births [12]. Antenatal attendance among pregnant women is high, with more than 95% attending at least once [13].

Ultrasound Equipment

Three portable SonoSite S180 machines equipped with curved 5-2MHz C60 transducers (SonoSite, Inc, Bothell, Washington) were purchased for image acquisition in GRAPHS. The S180 machine was initially developed as a portable point-of-care ultrasound machine for the US Department of Defense for use on the battlefield and, as such, offers durability and robustness atypical of many other machines.

Ultrasound Training

Ultrasound training occurred in several phases over 2 years as summarized in Table 1. We targeted midwives for ultrasound training rather than ultrasound technicians so that acquired skills could elevate obstetric care at participating hospitals. Phase 1 consisted of a 1-week introductory course in basic obstetric ultrasound that was developed and led by the GRAPHS obstetrician (BJW). All 15 midwives that provided antenatal care at either Kintampo North Municipal Hospital in Kintampo or Kintampo South District Hospital in Jema participated. Topics covered included assessment of viability, location of the pregnancy, placental location, plurality of the gestation, and a basic introduction to fetal biometry. Didactics were combined with supervised practice. A special emphasis was placed on how to turn on and use the ultrasound machines located at each hospital so that basic point-of-care ultrasound could be incorporated as needed when formal ultrasound at the hospital was unavailable. Participating midwives received a certificate of attendance in Basic Obstetric Ultrasound following this course of instruction.

A US-based registered diagnostic medical sonographer travelled to Kintampo to conduct Phase 2 of the ultrasound training during May 2012. A group of five midwives and one medical assistant from the two district hospitals were selected to undergo advanced training in this intensive hands-on course based on their performance in the first phase of training. Participants were instructed in the acquisition of fetal biometric measurements including first trimester measurements such as crown-rump length (CRL) and second trimester measurements such as biparietal diameter (BPD) and femur length (FL). Didactics emphasized identifying appropriate landmarks, ensuring adequate magnification of images, and placing calipers in the correct location. All training materials were developed by the study obstetrician using standard obstetric ultrasound textbooks. Sessions were devoted to practicing fetal biometry on volunteer subjects with immediate feedback and tips provided by the US sonographer (Figure 1). Additional practice was assigned upon identification of specific deficiencies.

Of the six midwives that underwent the intensive Phase 2 training, four became GRAPHS sonographers based on their aptitude, interest, and anticipated availability for the duration of the study. Additional supervised practice in fetal biometry for these four midwives (Phase 3) occurred through the Alliance for Maternal and Newborn Health Improvement Study (AMANHI; Clinical Trials Registration NCT01699945). In AMANHI, 1000 pregnant women underwent ultrasound examinations at less than 20 weeks in Kintampo. GRAPHS sonographers were paired with two US maternal-fetal medicine fellows to perform the AMANHI ultrasound scans. This afforded the GRAPHS sonographers the opportunity to solidify their scanning skills with an immediately available experienced teacher.

During the second half of 2013, all GRAPHS activities with the exception of the intervention were piloted in a cohort of 50 pregnant women. Ultrasound images obtained in the course of the pilot were saved in a de-identified form and transferred electronically to the GRAPHS obstetrician for remote review and feedback constituting Phase 4 of ultrasound training.

Figure 1. Hands-on ultrasound training of Ghanaian midwives by US-based registered diagnostic medical sonographer.

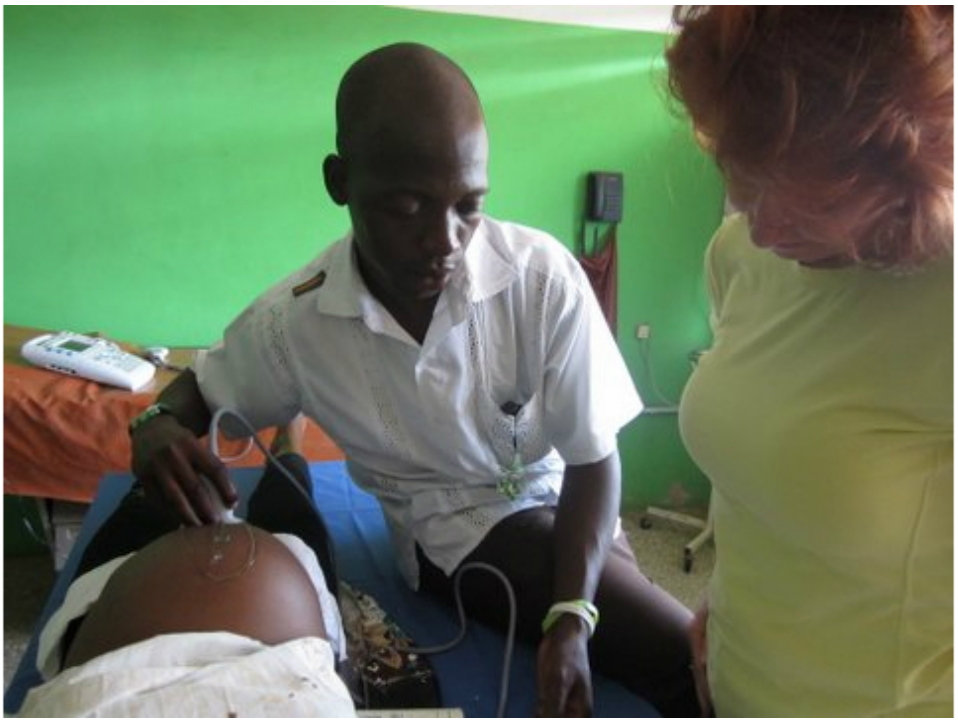


Table 1. Phases of ultrasound training in GRAPHS[a].

Phase of training	Trainer	Participants	Length of training	Topics covered
Phase 1	Trial obstetrician-perinatologist	All midwives (15) performing antenatal care at study district hospitals	1 week	Didactics: Basic obstetric ultrasound including assessment of viability, location of the pregnancy, placental location, plurality of the gestation, introduction to fetal biometry Hands-on: viability, pregnancy location, plurality, placental location
Phase 2	US-based registered diagnostic medical sonographer	5 midwives and 1 medical assistant, selected after Phase 1 for demonstration of ultrasound aptitude	2 weeks	Didactics: Focus on fetal biometry (CRL, BPD, FL[b]), appropriate landmark identification, adequate image magnification, correct caliper placement Hands-on: Fetal biometry all trimesters
Phase 3	US-based maternal-fetal medicine fellows	4 midwives selected after Phase 2 for aptitude and interest in ultrasound plus anticipated availability for trial duration	3 months	Hands-on: Fetal biometry <20 weeks gestation; practice scanning in 1000 subjects enrolled into separate maternal child health study
Phase 4	Trial obstetrician-perinatologist	4 study sonographers trained in Phase 3	6 months	Hands-on: Image acquisition of pilot GRAPHS participants; image review and written feedback by study obstetrician-perinatologist

[a]GRAPHS: Ghana Randomized Air Pollution Health Study

[b]CRL: crown-rump length; BPD: biparietal diameter; FL: femur length

Ultrasound Protocol in GRAPHS

Overview

GRAPHS employs resident field workers in all 35 clusters. These field workers identify pregnant women for potential trial participation. Interested subjects are referred to the Kintampo Municipal and Kintampo South District Hospitals for completion of screening.

Basic Overview Scan

At the screening visit, the study midwife equipped with a portable ultrasound machine, ultrasound transmission gel, tissue, as well as the ultrasound gestational age form performs the screening ultrasound examinations on all referred pregnant women. A screening patient identification number is generated by the hospital-based field worker and provided to the research midwife for entry into the ultrasound machine so no personal identifying information is recorded or stored with the ultrasound images. The entirety of the abdomen is scanned to confirm pregnancy, to identify the location, and to verify a single live fetus (Table 2). If the pregnancy is too early to be visible by ultrasound, warning signs of an ectopic gestation are reviewed and a follow-up ultrasound rescheduled in 1 to 2 weeks. If twins or higher order multiples are discovered or if the fetus is not alive, the scan is concluded, the woman is deemed ineligible for trial participation, and she is referred for antenatal care with a notation made of the scan's findings. Biometric measurements are obtained for those still eligible after this overview scan (described below). All women receive a printed ultrasound image of their baby.

Table 2. Components of the overview scan in GRAPHS[a].

Components of overview scan	Comments
Determine if subject pregnant	If no fetal pole visible within uterus, repeat ultrasound scheduled in 1-2 weeks; ectopic precautions reviewed
Establish location of pregnancy	If extrauterine location suspected, woman referred urgently for care at hospital
Determine plurality of gestation	Women carrying more than one fetus referred for routine antenatal care
Evaluate viability of pregnancy	If no fetal heart motion detected, repeat ultrasound scheduled if ≤10 weeks; if >10 weeks, referred to hospital for evaluation of potential missed abortion

[a]GRAPHS: Ghana Randomized Air Pollution Health Study

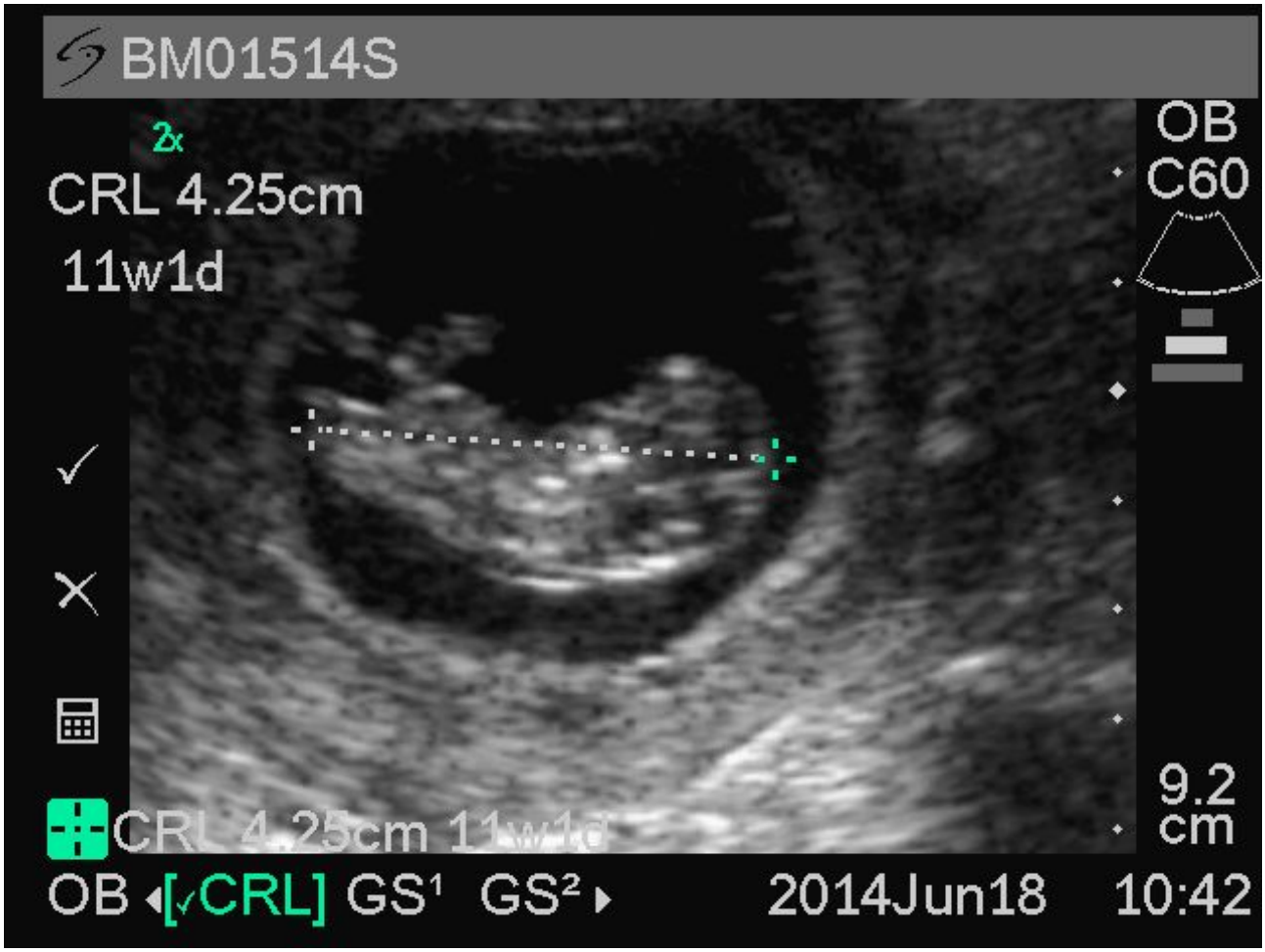
Fetal Biometry

First Trimester Biometry (<14 Weeks Gestation)

For first trimester scans, midwives are instructed to obtain CRL measurements for pregnancies less than 14 weeks. When both the crown and rump are visible on the screen in the approximate midsagital plane imaged horizontally on the ultrasound screen, the image is zoomed to occupy more than 50% of the screen and frozen. Calipers are placed at the exterior edge of the skull (the crown) and at the inferior aspect of the pelvic bones (the

rump). A sample GRAPHS CRL image is shown in Figure 2. The measured image is then saved to the portable ultrasound machine. A total of three CRL images are obtained. A summary report is generated by the ultrasound machine, which averages the three saved images to estimate an overall gestational age and corresponding estimated date of delivery. Both the measurements and the corresponding gestational age estimated by the machine software are recorded onto the Ultrasound Gestational Age Form. The machine-generated delivery date is considered the Working Estimated Delivery Date for the trial and is used to schedule all other trial activities and to anticipate the delivery.

Figure 2. Representative sample image of crown-rump length from Ghana Randomized Air Pollution and Health Study (GRAPHS) participant.



Second Trimester Biometry (14-24 Weeks Gestation)

For second trimester scans, midwives are instructed to obtain BPD and FL measurements for pregnancies 14 weeks gestation or more (Table 3). The abdominal circumference is not used in GRAPHS as the majority of subjects are screened prior to 20 weeks when this biometric measurement is less relevant for gestational age determination. For fetuses measuring 14 weeks by BPD, additional CRL images are obtained to help verify the gestational age. To obtain a quality BPD image, study midwives are instructed to identify the fetal head, orienting the transducer so that the skull is imaged side to side on the screen, is visible as a continuous echogenic line, and appears oval rather than round in shape. The thalamus (butterfly appearance) and the cavum septum pellucidum (looks like an equal sign) are then identified to find the appropriate plane and the image zoomed to occupy more than 50% of the screen prior to freezing. The calipers are then placed at the widest portion of the skull from outer-to-inner. The top caliper is placed in contact with the external surface of the echogenic skull line and the bottom caliper placed along the inner surface of the echogenic skull line on the lower portion of the image. The measurement line intersects the longitudinal axis of the skull at a right angle. A representative BPD image from GRAPHS is shown in Figure 3. The measured image is then saved to the portable ultrasound machine. A total of three BPD images are obtained.

To obtain a quality FL image, study midwives are instructed to scan axially from the fetal abdomen toward the fetal pelvis until the bladder is identified. The echogenic area closest to the skin outside the pelvis is the femur in cross-section. Turning 90 degrees on this echogenic circle elongates the femur into view. The transducer is oriented so that the full extent of the femur is visible side to side on the screen and appears straight rather than curved. Calipers are placed at the external edges of the femur bone without inclusion of the secondary ossification center. A representative image from GRAPHS is shown in Figure 4. By scanning through the abdomen to the pelvis rather than randomly looking for fetal bones, the femur rather than the humerus is identified. The measured image is then saved to the portable ultrasound machine. A total of three FL images are obtained.

The ultrasound machine averages the three BPD and three FL measurements to estimate an overall gestational age and corresponding estimated date of delivery. Both the measurements and the corresponding gestational age estimated by the software are recorded onto the Ultrasound Gestational Age Form**.** The machine-generated delivery date serves as the Working Estimated Delivery Date for the trial and is used to schedule all other trial activities and to anticipate the delivery. Menstrual dates are not used.

Figure 3. Representative sample of biparietal diameter from Ghana Randomized Air Pollution and Health Study (GRAPHS) participant.

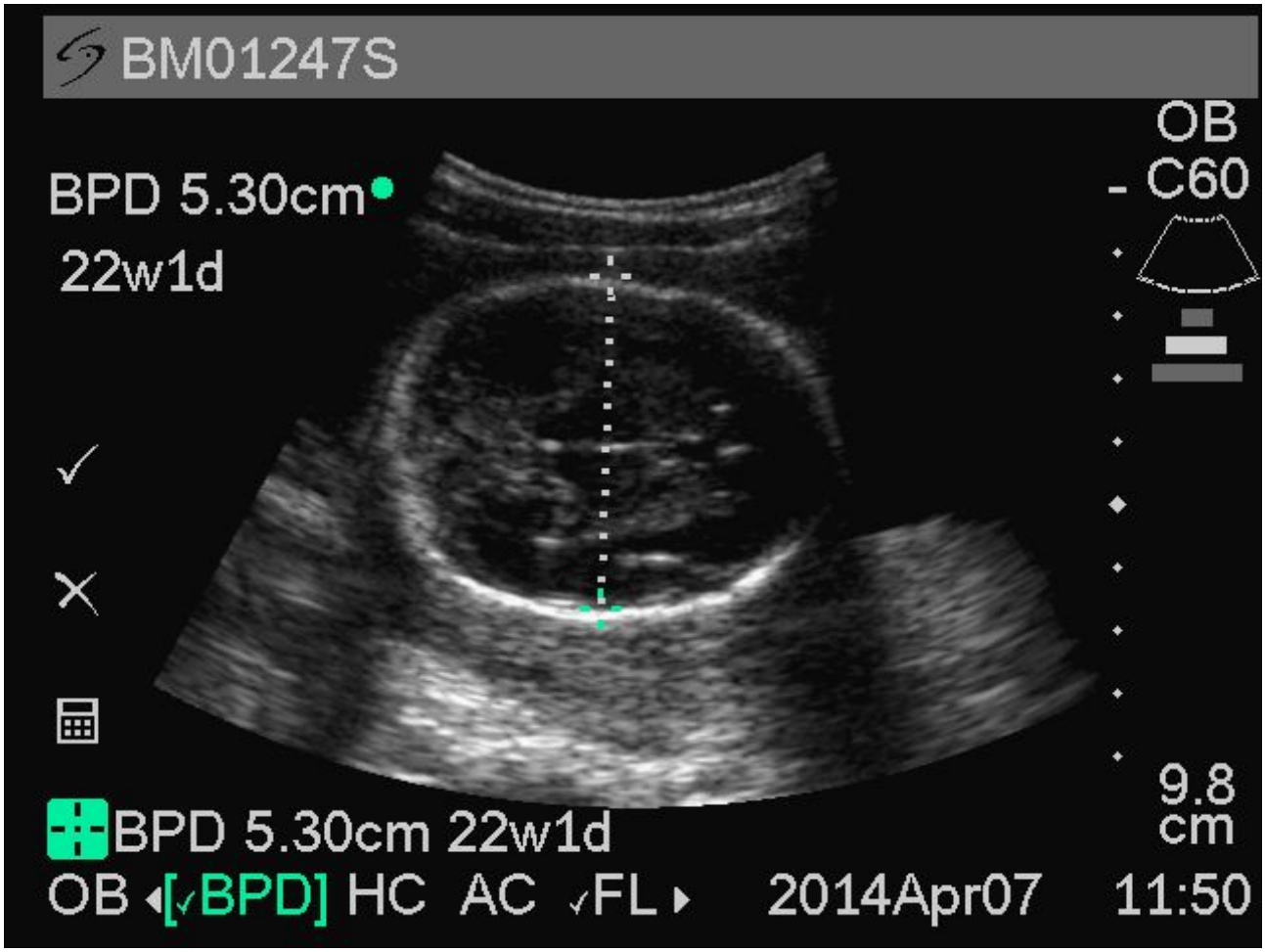
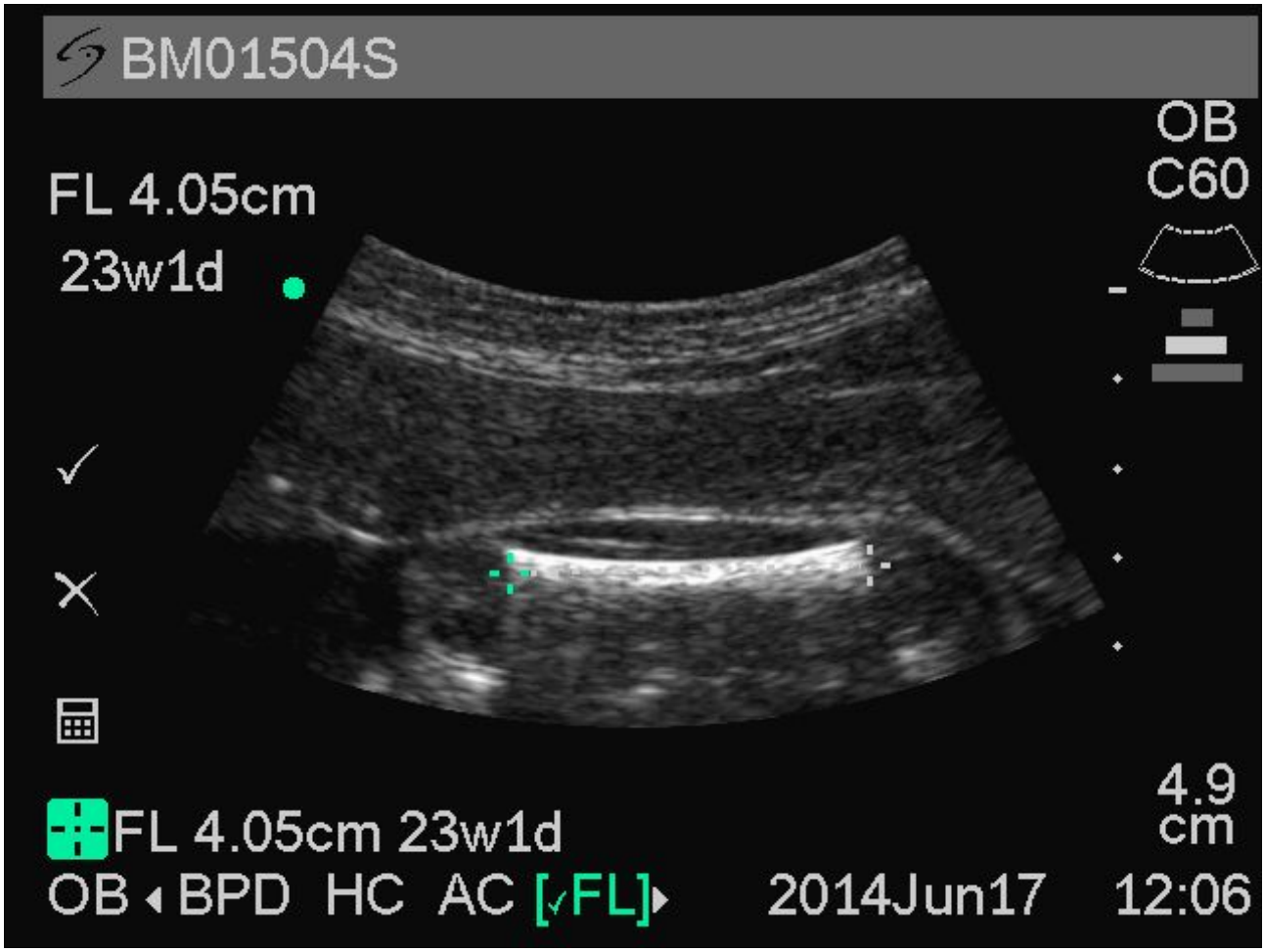


Figure 4. Representative sample image of femur length from Ghana Randomized Air Pollution and Health Study (GRAPHS) participant.



Transfer of Images

Images are transferred at the end of each day from the portable ultrasound machine to a password-protected computer in Kintampo and backed up to a secure server accessible by both the US- and Ghana-based researchers. Every 2 weeks, images are transferred electronically for review by the study obstetrician.

Gestational Age Assessment

The two best images for each biometric parameter are selected and averaged by the study obstetrician blinded to intervention arm. Should more than one image be deemed substandard, the single best image will be used. If all three images are unacceptable, research staff will be instructed to have the subject return for a second dating scan as soon as possible.

For pregnancies <14 weeks gestation, the average CRL measured is used to date the pregnancy (Table 3). Pregnancies measured in the 14th and 15th week are dated by the BPD only.

After 16 weeks, the selected BPD and FL measurements are then averaged to estimate the overall gestational age. The Trial Estimated Date of Delivery is then calculated using an electronic wheel, available as an app for download on mobile phones, and recorded in the Ultrasound Summary Report (Multimedia Appendix 1) [14]. As noted previously, the Working Estimated Date of Delivery will be used by research personnel to time research activities and plan for the anticipated birth. However, the Trial Estimated Date of Delivery will be the due date used in the analysis phase of GRAPHS to determine gestational age at enrollment and at delivery. This allows for removal of any images not meeting study standards.

While the ultrasound scan included in the GRAPHS trial is not intended as a screen for fetal anatomy, the study obstetrician reports any anomalies suspected in the images she reviews to the trial supervisor. These women are referred for formal ultrasound with notations as to the suspected anomaly. Similarly, any abnormalities suspected by the study midwives are transferred to the study obstetrician for more urgent review.

Table 3. Biometric parameter used to establish gestational age in GRAPHS[a].

Gestational age	Biometric parameter used to establish trial estimated delivery date
Less than 14 0/7 weeks gestation	CRL, average of two best images[c]
14 0/7 through 15 6/7 weeks gestation[b]	BPD, average of two best images[c]
16 0/7 weeks gestation and greater	BPD +FL, average of two best BPD and two best FL images[c]

[a]GRAPHS: Ghana Randomized Air Pollution Health Study.

[b]Research midwives are encouraged to obtain both CRL and BPD for pregnancies where gestational age appears to be in the 13-15 week range.

[c]Best images are determined by study obstetrician following review of all images.

Ongoing Quality Review

At the time of image review for gestational age assessment, the study obstetrician identifies any image quality concerns that require feedback. Such commentary has included reminders about appropriate zooming of images and gentle admonishments about precise caliper placement. One study sonographer's employment with the trial was terminated due to an inability to meet quality standards.

In addition to this informal immediate feedback, 5% of all subjects are selected randomly by the trial supervisor for formal quality review by the study obstetrician. Using a set of predefined criteria (Table 4), the scan is formally graded and recorded onto a MS Excel spreadsheet for the sonographer. A maximum of 7 points for each biometric parameter is awarded in the areas of correct magnification, correct plane identification, and correct caliper placements. These formal reviews are shared with the study midwives for performance improvement and kept as part of the study staff's employee file.

Table 4. Quality scorecard for components of fetal biometric measurements.

	Crown rump length	Biparietal diameter	Femur length
Quality criteria	1 point each[a]	1 point each[a]	1 point each[a]
Correct magnification			
	Good magnification (>50% of image)	Good magnification (>50% of image)	Good magnification (>50% of image)
	Neutral position (not hyperflexed or hyperextended)	--	--
Correct plane			
	Fetus horizontal (side to side on screen)	Skull is oval and visible throughout	Femur imaged side to side on screen
	Full extent of crown visible	Thalamus is visible	Only one bone in this portion of the extremity
	Full extent of rump visible	Skull side to side on screen	Upper femur measured
	--	--	Full extent of femur visualized (solid straight line)
Correct caliper placement			
	Crown caliper at exterior edge of skull	Calipers placed perpendicular to the long axis of the skull	Calipers placed at edge of echogenic bone (outer to outer)
	Rump caliper placed at lower spine	Top caliper placed on outer portion of skull	Secondary ossification centers not measured
	--	Bottom caliper placed on inner portion of skull	--
Total score	Maximum=7 points	Maximum=7 points	Maximum=7 points

[a]Half-points can be awarded.

Trial Ultrasound Examinations Through June 2014

Since launch of the trial, 1310 pregnant women have been screened for eligibility with an ultrasound examination through the end of June 2014. Of these scans, 146 women were found to be ineligible for the study based upon findings during the ultrasound. This included 91 women whose pregnancy was found to be greater than 24 weeks by fetal biometry, 38 women found not to be pregnant, 16 women carrying twins, and one woman with a nonviable fetus. In addition, 43 examinations were rescheduled as the pregnancy was too early to confirm and date by ultrasound. Of the 513 births that have occurred

among those enrolled in the trial, 3 sets of undiagnosed twins were born. One case of fetal hydrocephalus was discovered during Phase 2 of ultrasound training and a cystic hygroma identified in one of the first trimester screening scans.

Discussion

Principal Findings

We have described the extensive training and ongoing quality control program developed to ensure a high caliber of ultrasound examination in GRAPHS currently underway in in rural Ghana. The primary objective of the ultrasound training and continuing review is to optimize gestational age assessment in the trial. Accurate gestational age is required to target intervention deployment prior to 28 weeks and to evaluate whether the stove interventions impact rates of preterm birth (delivery <37 weeks) or small for gestational age (birth weight less than 10^{th} percentile), which are secondary outcomes of the study. Additional benefits of ultrasound at enrollment include verification of other trial inclusion criteria such as a singleton gestation, live fetus, intrauterine location, and comparability of study arms. We also anticipate that ultrasound examination and provision of a photo might increase satisfaction of the participating mother.

An equally compelling rationale underlying our decision to include ultrasound in GRAPHS was the training and capacity building this endeavor would bring to both the Kintampo Health Research Centre and the two hospitals where the ultrasounds are being performed. Ultrasound scans can reveal high-risk situations such as women carrying multiples or anomalous fetuses. Potentially life-threatening pregnancy complications such as ectopic gestation and placenta previa also can be identified. The benefits of identifying complicated pregnancies may be even greater compared to resourced settings as maternal and perinatal mortality and morbidity are much higher in low resource settings [15,16]. Ultrasound machines were previously available at both study hospitals but routinely used only during daytime hours and dependent upon physician or radiology technician availability. Prior to the GRAPHS ultrasound training, none of the midwives who provide the majority of antenatal care had been taught to use the machines or interpret the images. By orienting the group of midwives performing antenatal care at these hospitals to the use of the machine and basic obstetric scanning, it is our hope that point-of-care ultrasound will be embraced by the antenatal providers. Capacity is therefore built both for the research center and for the clinical community.

Several key points bear additional emphasis. Building ultrasound capacity was not an overnight phenomenon. This endeavor required a significant investment of time both by educators and trainees. Ultrasound skills cannot be taught in the classroom. While didactics are useful to identify target landmarks, hands-on practice is required. Obstetric scanning necessitates a spatial awareness and an ability to move a transducer in multiple planes as the fetus is mobile and not always in the same orientation. A certain amount of flexibility and fluidity on the part of the person scanning is required to follow the baby and find the correct planes.

Beyond acquisition of high quality images, accurate gestational age assessment requires a standardized protocol. Specifying a priori which biometric parameter will be used for pregnancy dating at different time points in gestation avoids any suggestion that findings were fit to study hypotheses. A single electronic wheel is being used for all GRAPHS participants as many paper-based pregnancy wheels vary in estimated delivery dates depending on the physical size of the wheel. Verification of the gestational age in GRAPHS is time intensive. The study obstetrician personally reviews every ultrasound image for GRAPHS participants to select those suitable for inclusion in dating the pregnancy. Her clinical career as a maternal-fetal medicine specialist has included the review of over 10,000 ultrasound examinations and she therefore brings considerable experience to the trial. These skills are being transferred over the course of the trial to the Ghanaian research team such that future research trials will have local capacity for gestational age assessment.

Conclusions

We have demonstrated that it is feasible to obtain ultrasound biometry for the accurate establishment of gestational age in a large field-based trial in rural West Africa, following a significant commitment of time, dedication, and resources to achieve and maintain high quality. We have presented our extensive ultrasound training program, quality control process, and gestational age assessment protocol from the GRAPHS trial in the anticipation that our experience may be of utility to others engaged in obstetric research in resource-limited settings.

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Conflicts of Interest

None declared.

Multimedia Appendix 1

Representative sample of Ultrasound Summary Report.

[PDF File (Adobe PDF File), 6KB - resprot_v3i4e77_app1.pdf]

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Abbreviations

AMANHI: Alliance for Maternal and Newborn Health Improvement Study
BPD: biparietal diameter
CRL: crown-rump length
FL: femur length
GRAPHS: Ghana Randomized Air Pollution Health Study
LPG: liquefied petroleum gas

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