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Agile development of a digital exposure treatment for youth with chronic musculoskeletal pain: protocol of a user-centred design approach and examination of feasibility and preliminary efficacy

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ABSTRACT

Introduction Chronic pain affects a significant number of children and impacts multiple domains including social, emotional and behavioural functioning, and negatively impacts family functioning. Roughly 5% of youth with chronic pain experience moderate to severe pain-related disability, with pain-related fear and avoidance of activities being identified as substantial barriers to treatment engagement. Evidence supports targeted psychological and physical interventions to address these barriers (eg, graded-exposure treatment), but accessibility to intervention is undermined by a shortage of services outside of urban areas, high treatment-related costs, and long provider waitlists; highlighting the need to develop digitally delivered behavioural intervention, using agile and iterative study designs that support rapid development and timely dissemination.

Methods and analysis This study seeks to develop an effective and scalable intervention for youth with chronic pain and their caregivers. This paper presents a user-centred protocol for the development and refinement of a digital exposure treatment for youth and caregivers, as well as the study design to examine feasibility and preliminary efficacy of the treatment using single-case experimental design (SCED). Assessments include daily diaries, completed from baseline and daily throughout the intervention (~6 weeks), and at 3-month follow-up, as well as self-report measures completed at baseline, end of intervention and 3-month follow-up. Primary outcomes include treatment satisfaction, treatment expectancy, adherence to daily dairies and functional disability. Secondary outcomes are pain-related fear and avoidance of activities, pain catastrophising and pain acceptance. We will present descriptive and model-based inference analyses, based on SCED reporting guidelines. We will calculate effect sizes for each individual on each outcome. We will examine mean treatment expectancy, credibility and satisfaction scores, and patient drop-out percentage.

Ethics and dissemination This study is approved by the Institutional Review Board at Stanford University (protocol #53323). Findings will be actively disseminated through peer-reviewed journals, conference presentations and social media.

Trial registration number NCT05079984.

STRENGTHS AND LIMITATIONS OF THIS STUDY

⇒ An iterative, agile, data-driven approach to development of digital interventions provides promise to support the rapid evaluation and implementation of digital tools.
⇒ User-centred design process (ie, youth with lived experiences of chronic pain and caregivers) allows for evaluation of comprehensiveness and acceptability of treatment content and structure directly from end-users.
⇒ Single-case experimental design supports the agile, iterative development of digital tools, allowing feasibility and preliminary efficacy to be examined at the individual (vs group) level.

INTRODUCTION

Chronic pain affects the lives of over a quarter of youth,[1–3] with societal costs exceeding US$19.5 billion dollars in the USA each year.[4] The negative impact of chronic pain is reported across many domains (eg, social, emotional and behavioural functioning), as well as family functioning.[5–7] Particularly, pain-related fear and avoidance of activities are known to significantly interfere with treatment engagement and negatively impact pain outcomes for youth.[8 9] This results in continued high healthcare utilisation without symptom improvement[10] and highlights the need for a treatment approach that directly targets pain-related fear and activity engagement as a pathway to enhance patient outcomes.[10–13]

Graded-exposure treatment (GET)[14] is a theory-driven,[15] individually tailored and evidence-based[16–18] behavioural intervention for individuals with chronic pain that targets pain-related impairment through exposure to previously feared and avoided activities. GET has robust effect sizes in adults[16] and has demonstrated similar outcomes among adolescents.[18–21] Despite evidence supporting early, targeted and integrated psychological and physiological interventions,[10] accessibility is undermined by a shortage of services, significant treatment-related cost4s and long provider waitlists.[4 22] These barriers underscore the critical need for continued development of innovative, digitally delivered interventions for youth, increasing the scale of these interventions.

Digital health represents a solution to access-to-care barriers[23–33] and outcomes of digital interventions for youth with chronic pain are similar to in vivo treatment.[34] However, only 28% of internet or smartphone application tools reach end-users and have meaningful adoption, leading to suboptimal healthcare innovation and significant research waste.[35] Digital health interventions developed within academia are theory-driven and evaluated scientifically,[36] but methods such as randomised controlled trials (RCTs) are time-consuming and costly, often preventing rapid dissemination and implementation. This highlights the inflexibility of the academic approach compared with industry development, which utilises repeated, rapid cycles of fine-tuning based on user feedback.[37] However, most digital health interventions developed within industry lack a clear theoretical framework,[38–40] evidence-based content[41–44] and systematic effectiveness testing.[45]

The mHealth Agile Development and Lifecycle model (figure 1)[46] provides a conceptual framework for the iterative, user-centred, rapid evaluation of sustainable, evidence-based digital solutions, bridging the gap between academia and industry. Using flexible research designs and methodologies to examine treatment effects can also support this effort. Single-case experimental design (SCED), for example, is an experimental method aimed at testing the effect of an intervention using a small number of patients (also termed N=1 trials),[47 48] using repeated measures, sequential randomised introduction of an intervention and method-specific data analysis.[48] Coupled together, SCED and the mHealth Agile Development and Lifecycle model provide the opportunity to rapidly and pragmatically develop and evaluate digital interventions in small cohorts.[49 50]

This paper presents the protocol for the development of a digital exposure intervention for youth with chronic musculoskeletal (MSK) pain and their caregivers using the mHealth Agile Development Lifecycle model and SCED methodology. To develop and refine a prototype of the youth and caregiver interventions, we will use an iterative, user-centred design process—interviewing with youth with lived experiences of chronic pain and caregivers to evaluate comprehensiveness and acceptability of treatment content and structure and subsequently the feasibility and preliminary efficacy of the interventions on improving outcomes for youth and their caregivers.

METHODS

Development of the digital solution will be based on a series of semistructured interviews with youth with lived experiences of chronic pain and their caregivers (phase 1). Once an acceptable version of the intervention is developed, feasibility and preliminary efficacy will be examined using SCED with a sample of naïve end-users (youth and caregivers; phase 2). This study is approved by the Institutional Review Board at Stanford University

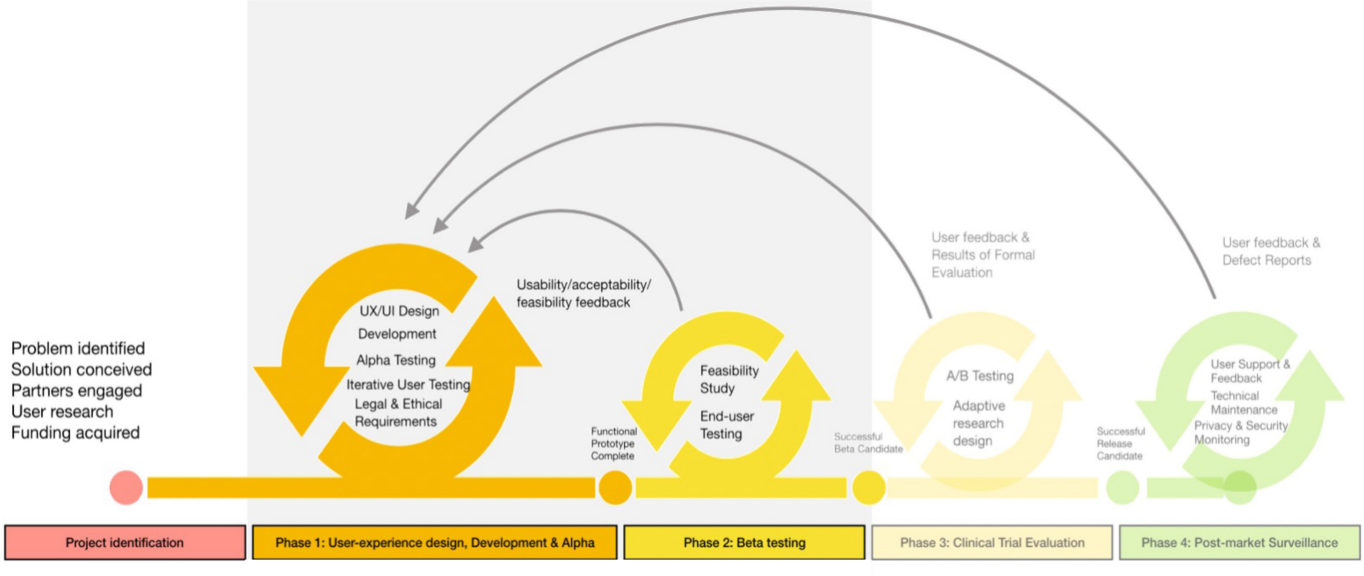

Figure 1 Phases of development of iGET Living situated within the mHealth Agile development and lifecycle model.

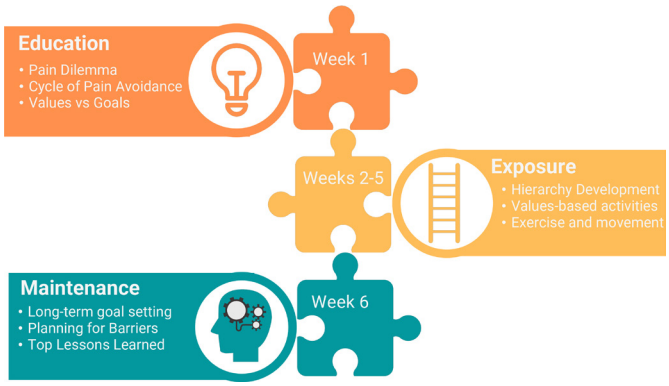

Figure 2 Core components of iGET Living to support behaviour change.

(protocol #53323) and registered at ClinicalTrials.gov (NCT05079984).

Recruitment and participants

All patients presenting to the Paediatric Pain Management Clinic at Stanford Children's Health complete Peds-CHOIR[51] and can indicate if they would like to participate in research. More than 650 patients have expressed interest in research, with~300 new patient evaluations conducted each year. We will leverage this registry for recruitment. We will also recruit participants broadly (via professional listservs with providers who work with youth with chronic pain, chronic pain online support groups, etc). Patients who meet eligibility criteria and express interest in participation via a brief online screening form will receive a recruitment phone call from a research assistant to introduce the study and confirm eligibility.

Phase 1

User-centred development. Youth (n=15) will be 10–18 years old, have a diagnosis of chronic (>3 months)[52] MSK pain (ie, pain in the bones, joints, ligaments, tendons or muscles), be English literate, and have access to a computer, smartphone or tablet with internet connection. Caregivers (ie, any adult, legal guardian who cares for the youth; n=15) will be self-identified caregivers of youth with chronic pain. Caregiver–child dyads are preferrable but not required in this phase.

Phase 2

Feasibility and preliminary efficacy. Youth (n=20) ages 10–18 years and one of their caregivers (n=20) will be recruited. Youth will have a diagnosis of chronic (>3 months) MSK pain, have moderate to high pain-related disability (Functional Disability Inventory (FDI)[53]≥13), be English literate, and have access to a computer, smartphone or tablet with internet connection. Youth will be ineligible to participate if they demonstrate significant cognitive impairment (eg, brain or significant medical or psychiatric problems that would interfere with treatment (eg, psychosis, suicidality) or have engaged in pain psychology treatment (eg, cognitive-behavioural therapy, acceptance and commitment therapy, exposure therapy) 6 months prior to enrolment. iGET Living: Youth Intervention

A prototype of the digital exposure intervention, iGET Living, for youth was developed from an in-person graded-exposure intervention for youth (GET Living),[17] which was recently evaluated in a two-arm RCT (please see Simons *et al*, 2020 for an in-depth description of the in-person intervention; Simons *et al*, in preparation). The prototype of iGET Living is built on a secure flexible platform that can be used on any device (BASS4; https://ki.se/en/research/internet-delivered-interventions). BASS4 is available through the Karolinska Institutet e-Health Core Facility which provides maintenance and technical support and has been successfully used for other internet-based behavioural interventions with adolescents and adults.[54–56] iGET Living extracts fundamental concepts from in-person GET Living, such as focusing on pain-related fear and avoidance as key targeted mechanisms to increase functioning, as well as focusing on pain willingness and values-based actions in the context of exposures (figure 2). Different from in-person GET Living, which is delivered twice weekly and jointly by a physical therapist and psychologist, iGET Living has been restructured for daily engagement and is meant to be mostly self-guided. iGET Living is structured to be completed in ~6 weeks, with content organised into 30 brief (5–15 min daily engagement) modules, with the goal that patients complete a minimum of five sessions per week. Each patient is assigned a therapist (ie, licensed psychologist with experience treating youth with chronic pain, clinical psychology trainee) with whom they are able to communicate with during the intervention via a messaging functioning within the platform. Therapists are also able to see the patient's response to activities completed in the modules and can provide feedback. At the end of each week, the therapist sends a message to the patient, reflecting on their progress and providing support for any barriers to meeting goals. Messages are predrafted but tailored to the individual patient prior to deployment. The structure of the protocol was modelling after an ACT-based smartphone application for adults with chronic pain.[57]

iGET living: caregiver intervention

During in-person GET Living, caregivers are present for every treatment session with their child and received three, one-on-one sessions with the pain psychologist. We will extract fundamental concepts from the caregiver sessions, including education and a focus on caregiver distress in the context of their child's pain, for the digital intervention. We will use data from user-centred interviews to inform the structure and duration of the caregiver intervention.

Treatment components

Education, goal setting, and activity hierarchy development

The first six modules orient the user to the structure of the intervention and provide the rationale for exposure.

These sessions also provide education and training for skills youth will put in to practice in the following weeks, such as introduction to the 'Pain Dilemma' (the conceptualisation of the dysfunctional behavioural strategies, motivational interviewing towards focus on improvement of function vs pain elimination),[58] the pain-avoidance cycle, values clarification and goal setting, education on exposure and activity hierarchy development.

Enhancing function through achievable goals and exposures

In these modules, patients are guided through values clarification exercises for various domains (ie, friends/friendship, family, school and hobbies) and setting values-based treatment goals to support movement towards increased function vs pain reduction. Activity exposures are designed to support the youth in engaging in activities that are feared or avoided due to the presence of pain. These activity exposures can be related to their values-based goal for the week. For example, having clarified that part of being a good friend means communicating with them regularly, a goal could be to have three phone calls during the week or text at least once daily. In support of this goal, one of the activity exposures could be to engaging in the phone call. Activity exposures can also be related to a physical activity (eg, walking for 15 min) or another activity that the youth has identified as something they want to or need to be doing (eg, helping with chores). Each week ends with a self-reflection on progress and barriers to meeting the goal or engaging in the exposure as well as progress made.

Planning for barriers and long-term goal setting

Towards the end of the intervention, patients engage in setting long-term goals (ie, goals to work towards in the coming 3 months). Time is also spent identifying potential barriers for each goal and developing 1–2 potential solutions for overcoming those barriers. Patients also engage in thought-challenging exercises focused on negative automatic thoughts that could arise when met with a barrier or set back and are supported in generating alternative thoughts to support continued progress forward. The last module is an exercise called 'Top Lessons Learnt' where patients list the key take-aways learnt during the intervention and skills they want to remember moving forward.

Caregiver content

Similar to youth, caregivers will receive education related to pain-related fear and avoidance as key mechanisms linked to function, as well as focusing on pain willingness and values-based actions in the context of exposures to enhance function. Content will also focus on caregiver distress in the context of their child's pain and behaviour change: strategies for promoting activity engagement, reacting vs responding to child pain symptoms, and the concept of rescuing versus riding it out when the child is in distress. We will also assess caregiver experiences (if any) with pain psychology interventions to date, including needs and wants for an ideal intervention. Taken together, these data will inform the development of a caregiver digital prototype.

Phase 1: user-centred development

Interview sessions with youth and caregivers will be conducted by a member of the research team via Zoom, an online videoconference platform for healthcare, and will use best practices for qualitative and cognitive interviewing for research.[32 33 59] Consent and assent (see online supplemental materials 1; 2) will be obtained through the secure, web-based application REDCap (Research Electronic Data Capture). Each participant will be given a log-in and password allowing them to access the BASS4 platform where they will be able to view and pilot various functions within the platform (eg, audio files, animations, completing activities). While viewing treatment content, participants will be prompted to think aloud about likes, dislikes, and difficulties with the content. Likert scale-rated and open-ended questions assessing acceptability, ease of use, comprehensibility, and suggestions for improvement will also be administered. Items will be administered after each module, as well as at the conclusion of viewing all content.

All interviews will be recorded and transcribed, and data will be entered and stored on REDCap. Videos will be stored on a secure cloud-based platform safe for personal health information and accessed only by the research team. Interview sessions will continue until data saturation is reached (ie, no data/feedback are generated that have not already been categorised). Once data saturation is reached, all usability data from a single iterative cycle will be considered to have been identified[60 61] and the content of the interventions will be modified accordingly. Review of the literature and previously conducted usability testing studies indicates that prototype refinement is typically achieved within 2–3 cycles of testing, with 4–5 participants in each cycle.[60 61] Therefore, we aim to conduct three iterative cycles of user-centred interviews.

Phase 2: feasibility and preliminary efficacy

Participants will complete a remote baseline study visit with a research assistant via Zoom. Prior to the baseline visit, a wearable Actigraph[62] will be mailed to each youth participant via US postal service priority mail, and they will be instructed to begin wearing the device immediately following their remote baseline visit until completion of the intervention.

Baseline

Consent, assent and self-report measures will be collected via REDCap from youth and caregivers. Youth will complete a brief biomechanical assessment of walk speed and balance, which will be recorded and analysed using OpenPose.[63] Youth and caregivers also begin completing daily diaries at this time and are randomly assigned to a baseline period of 10–21 days (in line with SCED methodology for staggered introduction of the intervention

among participants). Following their baseline period, the patient begins the intervention. On the day they are scheduled to begin the intervention, the participant will be provided with a username, password and hyperlink used to access the BASS platform.

Intervention

Patients are encouraged to engage with the programme daily but we will use data from user-centred interviews to better inform expectations for frequency of engagement. Each daily module should take no more than 15 min to complete.

Completion of Treatment

At the end of treatment (~6 weeks), the patient will attend a discharge visit via Zoom. Similar to the baseline visit, participants will complete self-report measures via REDCap and youth will also complete the biomechanical assessment. Participants will also complete an exit interview assessing their experience of the intervention. Daily diaries are discontinued at this time and families will be provided with instructions and materials for returning the wearable physical activity tracker.

Follow-up

All non-daily assessments will be administered to youth and caregivers again at 3-month follow-up. Youth and caregivers will also complete the daily diaries for a 7-day period.

Extensive support will also be provided during the onboarding phase to ensure families are able to access the intervention. If a participant displays difficulty with daily engagement (2+ days no progress), a member of the research team will reach out to the family via the messaging feature in the platform and problem-solve barriers to engagement. During consent procedures, participants will be informed of their right to withdraw from the study at any time without penalty. As the intervention is self-guided and digital, we do not anticipate a scenario in which we would need to discontinue participation.

Assessment of outcomes

All self-report assessments and daily diaries will be completed online through REDCap.

User-centred development outcomes

Youth and caregivers will complete demographic and medical history (described below) and will undergo a series of qualitative interviews (up to three 2-hour sessions, for a total of 6 hours maximum time) during the viewing of the intervention content. The qualitative interview is a semistructured interview developed by this research team to elicit qualitative and quantitative feedback that will be analysed and used to modify the intervention content.

The primary and secondary outcomes described below will be collected during aim 2 from youth and caregivers. Nondaily measures will be completed at baseline, discharge and 3-month follow-up. Daily diaries will be delivered to the adolescent to assess outcomes within the last 24 hours from the start of the baseline phase to discharge and will also be collected for 7 days at 3-month follow-up (table 1).

Feasibility and preliminary efficacy outcomes

Implementation outcomes

Treatment satisfaction (youth and caregiver)

Treatment satisfaction will be evaluated using the Pain Service Satisfaction Test (PSST)[64] at the end of treatment. The PSST consists of 22 items and assesses the individuals experience with pain clinic services and will be slightly modified to fit with the current intervention.[17]

Treatment expectancy (youth and caregiver)

Expectations for treatment effectiveness will be assessed using the Credibility/Effectiveness Questionnaire (CEQ).[65] The CEQ is composed of six items rated on 1–9 or 0%–100% scale, depending on the item, and assesses expectations related to the effects of the current treatment across two subscales: credibility and expectancy. Credibility is calculated by taking a mean of items 1–3. For expectancy, we will use the expectancy item that asks patients to rate (0%–100%), 'By the end of [treatment), how much improvement do you think will occur?'[66]

Treatment adherence (youth and caregiver)

Adherence and retention will be assessed by examining adherence/completion of daily diaries, per cent of patients who drop out prior to treatment completion and percentage of treatment modules completed.

Efficacy outcomes

Functional Impairment (youth)

The FDI[53] will be used to assess functional impairment in youth. The FDI is a 15-item self-report measure assessing perceived difficulty of performing activities across contexts (home, school, social and physical). Items are rated on a 5-point Likert scale ranging from 0 (no trouble) to 4 (impossible). Items are summed to create a total score, with higher scores indicating greater disability. Scores on the FDI can be interpreted using established clinical reference points: 1–12=no/minimal disability, 13–29=moderate disability and 30–60=severe disability.[67]

Pain-related fear and avoidance (youth and caregiver)

The Fear of Pain Questionnaire, child report short form [68] is a 10-item measure that assesses pain-related fear and avoidance of activities. Items are rated on a 0 (strongly disagree) to 4 (strongly agree) Likert scale. Items are summed to create a total score for each subscale: Fear of Pain (four items) and Avoidance of Activities (six items). Higher scores indicate higher levels of pain-related fear and avoidance. The Parent Fear of Pain Questionnaire (PFOPQ)[7] assesses caregiver's fear and avoidance behaviours associated with their child's pain experiences. We will use a newly developed short form (PFOPQ-SF) which contains nine items assessed on a 5-point Likert scale (0=strongly disagree to 4=strongly agree). Items are summed to create a total score for each subscale: Fear

Table 1 List of measures

Outcomes and correlates	Questionnaires and tests	DD	Full version
Phase 1			
Acceptability	Qualitative interview+		
Phase 2: Feasibility			
Treatment satisfaction	Treatment satisfaction[64]; mean score ≥40 of 60; satisfied to very satisfied		END, FU
Treatment acceptability	Treatment expectancy and credibility[65]; % drop-out; Feedback from stakeholder interview (eg, patients, parents, clinicians)		BAS, END, FU
Treatment adherence	% adherence to daily diary, modules completed, # of messages to the therapist		END
Phase 2: Examination of effectiveness			
Primary Outcome: Functional Disability			
Functional disability	Functional Disability Inventory[53]	x	BAS, END, FU
Secondary outcomes: Pain-related distress and behaviour			
Pain-related distress	Fear of Pain Questionnaire, child report, short form[68]; Parent Fear of Pain Questionnaire[7]; Pain Catastrophising Scale-Child and Parent[80 6]	x	BAS, END, FU
Psychological Flexibility	Chronic Pain Acceptance Questionnaire for Adolescents[81]; Parent Psychological Flexibility Questionnaire[82]	x	BAS, END, FU
Parent Protective Behaviours	Adult Responses to Child's Symptoms[83]	x	BAS, END, FU
Correlates			
Pain Severity	Numerical Rating Scale[69]	x	BAS, END, FU
Medical History	Onset, location, duration, course, intensity of pain, current medications		BAS
Demographics	Age, gender, race, ethinicity, school grade, number of pain-related absenses, annual household income, parent hours worked in a week, parent days of work missed due to child's pain, zip code		BAS
Exploratory			
Biomechanics	Gait (stride length, velocity)[84]; dynamic postural control[85]		BAS, END
Physical Activity	Daily mean and peak activity via wearable device	x	

+, during viewing of content; BAS, baseline; DD, daily diary; END, discharge; FU, 3-month follow-up; TX, once weekly, during treatment.

of Pain and Movement, Avoidance of Activities, and Fear of School. Higher scores indicate higher levels of pain-related fear and avoidance.

Pain Catastrophising

The Pain Catastrophising Scale-C (PCS-C)[67] assesses negative pain-related cognitions. The PCS-C consists of 13-items rated on a 5-point Likert scale ranging from 0 (not at all true) to 4 (very true). Items are summed to create a total score, with higher scores indicating higher levels of catastrophic thinking. The Pain Catastrophising Scale-Parent Version (PCS-P)[6] assesses caregivers' negative cognitions associated with their child's pain. The PCS-P is composed of 13-itmes rated on a 5-point Likert scale ranging from 0 (not at all true) to 4 (very true). Items are summed to create a total score, with higher scores indicating higher levels of catastrophic thinking.

Pain acceptance

The Chronic Pain Acceptance Questionnaire for Adolescents- short form (CPAQ-A8)[68] will be used to assess acceptance of pain in youth. The CPAQ-A8 consists of eight items rated on a 5-point Likert scale ranging from 0 (never true) to 4 (always true) and examines pain acceptance across two subscales: activity engagement (four items) and pain willingness (four items). Items are summed to create a total score for each subscale, with higher scores indicating higher levels of activity engagement and pain willingness. The Parent Psychological Flexibility Questionnaire [69] is a 10-item questionnaire assessing caregiver's ability to accept their own distress and respond adaptively and flexibility to their child's pain. Items are rated on a 7-point Likert scale ranging from 0 (never true) to 6 (always true). Higher scores indicate greater caregiver psychological flexibility.

Caregiver Protective Responses

The Adult Responses to Children's Symptoms[70] is a self-report measure assessing caregiver behavioural responses to children's pain behaviours. We will use the Protect subscale (13 items), which examines protective caregiver behaviours such as limiting chores or other activities and

providing special attention in the context of pain symptoms. Items are rated on a 5-point Likert scale ranging from 0 (never) to 4 (always). A mean score is computed (ranging from 0 to 4), with higher scores indicating greater protective behaviours.

Covariates

Demographics and Medical History

Youth will self-report demographic variables, such as age, gender, race, ethnicity and current year in school. Caregivers will self-report age, gender, race, ethnicity, number of school days missed due to pain for their child, annual household income, caregiver labour force status, hours worked in a week and number of missed work-days due to their child's pain. Data related to onset, location, duration, course, intensity of pain symptoms and current medications will be assessed via self-report from youth and caregivers. These data will be collected at baseline.

Pain severity

Youth will provide their average pain rating for the past 24-hours on a standard Numerical Rating Scale[69] ranging from 0 (no pain) to 10 (worst possible pain) on a daily basis from baseline to end of treatment.[70 71] Average pain ratings will be calculated for 7 days prior to the first treatment session (baseline average pain) and 7 days after the end of treatment (average pain over the preceding week).

Exploratory

Biomechanical functioning

Assessment of physical function will include stride length and walk pace. These data will be collected via video and analysed using appropriate software (eg, OpenPose.[72] These data will be collected at baseline and discharge.

Daily activity level

We will collect daily physical activity data via a wearable physical activity tracker from youth while they are enrolled in the intervention.

Youth and caregiver daily diaries

The daily surveys for youth and caregivers will be used during phase 2. Youth diaries will assess engagement in values-based activities, self-efficacy, function, notable events and sleep. Caregiver diaries will assess their distress and behaviours in the context of their child's pain (eg, fear, catastrophising, avoidance), as well as personal/familial stressors. Items will be rated on a VAS ranging from 0 (strongly disagree) to 10 (strongly agree) and are administered in a random order each day to mitigate habitual responding. Youth and caregivers will receive a daily text message with a hyperlink to their daily survey. Participants can designate when during the day they prefer to receive the survey. Survey link becomes inactive after 24 hours of deployment.

Data analysis

User-centred development analyses

Qualitative data extraction

All videorecorded interviews will be transcribed verbatim. Transcripts (audio and typed feedback) will be uploaded into NVivo V.12. Framework matrix analysis will categorise emergent themes, yielding specific, recurring information. Modifications to content will be made after each iterative cycle (n=5) of testing.

Feasibility and preliminary efficacy analyses

Feasibility

We will examine mean treatment expectancy, credibility and satisfaction scores, and patient drop-out percentage (total N=20 for youth; N=20 for caregivers). For adherence, we will examine mean adolescent and parent daily diary completion and treatment module completion. A participant will be considered adherent if: ≥80% of modules completed, ≤20% attrition rate and ≥80% daily diary adherence.

Efficacy

Daily diaries: We will present descriptive analyses, based on single-case reporting guidelines[73] and recommendations for reporting on several replicated SCED cases.[74 75] Using the Shiny app for single-case data analysis,[76] we will calculate effect sizes for each individual on each outcome comparing phases (AB, BC, AC, where A=baseline, B=treatment, C=3-month follow-up) using the non-overlap of all pairs test statistic. Non-daily measures: Criterion of 30% improvement from baseline will be considered clinically significant change.[18]

Sample size and power analysis

Phase 1: user-centred development

Existing literature and previously conducted usability testing studies indicate that prototype refinement is typically achieved within 2–3 iterative cycles of user-centred interviews. Additionally, data saturation can usually be reached with samples as small as 5–7 participants per usability cycle.[77] Therefore, we aim to recruit 15 adolescents ages 10–18 years for up to three cycles of usability testing.

Phase 2: feasibility and preliminary efficacy

As SCED uses individual level (vs group level) analyses, power and sample size considerations do not apply. The design of this study was based on previous work done in adults (n=6[16]; n=8[78 79]) with chronic pain applying the same treatment approach and study design. We aim to recruit 20 participants for phase 2 to ensure adequate sample size and sufficient feasibility data.

Monitoring

This trial is monitored by Navitas Clinical Research, the executive secretary of the National Institute of Arthritis and Musculoskeletal and Skin Diseases (NIAMS). A safety monitoring committee of three experts, approved by NIAMS, will meet quarterly to review overall subject

enrolment status, accrual, adherence, protocol deviations and adverse events.

Trial status

This study was prospectively registered on ClinicalTrials.gov with the US National Library of Medicine on 10 April 2021 (NCT05079984). Participant recruitment for phase 1 (user-centred development) began March 2022 and is expected to be complete September 2022. We anticipate recruiting for phase 2 (feasibility and preliminary efficacy) from January 2023 to January 2024, with data collection ceasing approximately 6 months later. See online supplemental materials 3 for study timeline.

Patient and public involvement

Patients and caregivers will be directly involved in development of the intervention, including but not limited to, content, structure, time and frequency of engagement.

DISCUSSION

Exposure treatment is a promising intervention for youth with chronic pain, targeting both psychological and physical functioning. Digitalisation of effective interventions is crucial to increase reach and reduce care barriers. Further, engaging individuals with lived experiences in the development process is critical for treatment efficacy, and to enhance broad adoption by end users. This study presents the protocol for the development of a digital exposure treatment for youth with chronic pain. We aim to take a user-centred, codesign approach to the development of the intervention. Further, this study represents the first examination of feasibility and efficacy of a digital exposure-based treatment in this population using an SCED methodology. Future work will focus on the development of a companion caregiver intervention, which will highlight skills learnt by their child in the youth intervention while also targeting caregiver distress and behaviour within the context of their child's pain experience.

Ethics and dissemination

This study is approved by the Institutional Review Board at Stanford University (protocol #53323). All eligible participants will be informed of study procedures and will sign informed consent and assent forms if they agree to participate. Participants will be informed of their right to withdraw at any time without penalty. Each participant will receive a unique study ID code, ensuring anonymity.

Information about the project (eg, recruiting, enrolling) can be found on LEH's faculty website at Stanford University. The general outline of the project has been presented at one international conference. Following the publication of this study protocol paper, we aim to publish a number of peer-reviewed manuscripts. Any protocol modifications will be communicated. Results will also be disseminated at national and international conferences.

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Contributors LEH, LS, RW, JS and BD were involved in the conception and design of this project. LEH acquired and received the funding. RW, JS and BD contributed specific input related to user-centred development and implementation. LS and RW provided experience on single-case experimental design aspects of this project. SNW, ARVO, EC and NJ are research team members directly involved with carrying out study aims. LEH drafted the manuscript, and all authors revised the manuscript. All authors gave final approval and agreed to be accountable for all aspects of the manuscript.

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