BMJ Open is committed to open peer review. As part of this commitment we make the peer review history of every article we publish publicly available.

When an article is published we post the peer reviewers' comments and the authors' responses online. We also post the versions of the paper that were used during peer review. These are the versions that the peer review comments apply to.

The versions of the paper that follow are the versions that were submitted during the peer review process. They are not the versions of record or the final published versions. They should not be cited or distributed as the published version of this manuscript.

BMJ Open is an open access journal and the full, final, typeset and author-corrected version of record of the manuscript is available on our site with no access controls, subscription charges or pay-per-view fees (http://bmjopen.bmj.com).

If you have any questions on BMJ Open's open peer review process please email info.bmjopen@bmj.com

BMJ Open

Perspectives on Adapting a Mobile Application for Pain Self-Management in Neurofibromatosis Type 1: Recommendations of Individuals Living with Neurofibromatosis Type 1 and Pain Management Experts

Journal:	*BMJ Open*
Manuscript ID	bmjopen-2021-056692
Article Type:	Original research
Date Submitted by the Author:	25-Aug-2021
Complete List of Authors:	Grau, Lauretta; Yale University Yale School of Public Health, Department of Epidemiology of Microbial Diseases Larkin, Kaitlyn; Yale School of Medicine, Department of Psychiatry Lalloo, Chitra; The Hospital for Sick Children, Child Health Evaluative Sciences; University of Toronto, Institute of Health Policy, Management & Evaluation Stinson, Jennifer; Toronto SickKids, Child Health Evaluative Sciences; University of Toronto, Institute of Health Policy, Management & Evaluation Zempsky, William; University of Connecticut School of Medicine, Department of Pediatrics and Nursing; Connecticut Children's Medical Center, Division of Pain and Palliative Care Ball, Samuel; Yale School of Medicine, Department of Psychiatry Buono, Frank; Yale School of Medicine, Department of Psychiatry
Keywords:	COMPLEMENTARY MEDICINE, Adult neurology < NEUROLOGY, Neurological pain < NEUROLOGY, Neurogenetics < NEUROLOGY, PAIN MANAGEMENT, QUALITATIVE RESEARCH

SCHOLARONE™
Manuscripts

I, the Submitting Author has the right to grant and does grant on behalf of all authors of the Work (as defined in the below author licence), an exclusive licence and/or a non-exclusive licence for contributions from authors who are: i) UK Crown employees; ii) where BMJ has agreed a CC-BY licence shall apply, and/or iii) in accordance with the terms applicable for US Federal Government officers or employees acting as part of their official duties; on a worldwide, perpetual, irrevocable, royalty-free basis to BMJ Publishing Group Ltd ("BMJ") its licensees and where the relevant Journal is co-owned by BMJ to the co-owners of the Journal, to publish the Work in this journal and any other BMJ products and to exploit all rights, as set out in our licence.

The Submitting Author accepts and understands that any supply made under these terms is made by BMJ to the Submitting Author unless you are acting as an employee on behalf of your employer or a postgraduate student of an affiliated institution which is paying any applicable article publishing charge ("APC") for Open Access articles. Where the Submitting Author wishes to make the Work available on an Open Access basis (and intends to pay the relevant APC), the terms of reuse of such Open Access shall be governed by a Creative Commons licence – details of these licences and which Creative Commons licence will apply to this Work are set out in our licence referred to above.

Other than as permitted in any relevant BMJ Author's Self Archiving Policies, I confirm this Work has not been accepted for publication elsewhere, is not being considered for publication elsewhere and does not duplicate material already published. I confirm all authors consent to publication of this Work and authorise the granting of this licence.

Perspectives on Adapting a Mobile Application for Pain Self-Management in Neurofibromatosis Type 1: Recommendations of Individuals Living with Neurofibromatosis Type 1 and Pain Management Experts

Lauretta E. Grau, PhD[1], Kaitlyn Larkin, BA[2], Chitra Lalloo, PhD[3,4], Jennifer N. Stinson, RN, PhD[3,4], William T. Zempsky, MD[5,6], Samuel A. Ball PhD[2], and Frank D. Buono, PhD[2]

[1] Department of Epidemiology of Microbial Diseases, Yale School of Public Health, New Haven, CT USA
[2] Department of Psychiatry, Yale School of Medicine, New Haven, CT USA
[3] Child Health Evaluative Sciences, The Hospital for Sick Children, Toronto, Ontario
[4] Institute of Health Policy, Management & Evaluation, University of Toronto, Toronto, Ontario
[5] Department of Pediatrics and Nursing, University of Connecticut, Hartford, CT, USA
[6] Division of Pain and Palliative Medicine, Connecticut Children's Medical Center, Hartford, CT, USA

Corresponding Author:
Lauretta E. Grau, PhD
Yale School of Public Health
PO Box 208034
New Haven, CT 06520
E: lauretta.grau@yale.edu

Key words: COMPLEMENTARY MEDICINE, Adult Neurology, Neurological pain, Neurogenetics, PAIN MANAGEMENT, QUALITATIVE RESEARCH

Word count: 3863

ABSTRACT

Objectives: Neurofibromatosis Type 1 (NF1) is a genetic disorder in which chronic pain commonly occurs. The study sought to understand the needs of individuals with NF1 and pain management experts when adapting a pain self-management mobile health application for individuals with NF1. **Design:** We conducted a series of online, audio-recorded focus groups that were then thematically analyzed. **Setting:** Online focus groups with adults with NF1 currently residing in the U.S. **Participants:** Two types of participants were included: Individuals with NF1 (n = 32 across six focus groups) and pain management experts (n = 10 across three focus groups). **Results:** Six themes across two levels were identified. The Individual level included Lifestyle, Reasons for Using the Mobile App, and Concerns regarding its Use. The Application level included Desired Content, Desired Features, and Format Considerations. Findings included recommendations to grant free access to the application and include a community support feature for individuals to relate and validate one another's experience with pain from NF1. In addition, participants noted the importance of providing clear instructions on navigating the app, the use of an upbeat, hopeful tone, and appropriate visuals. **Conclusions:** Both participant groups endorsed the use of iCanCope as an NF1 pain self-management mobile application. Differences between groups was noted regarding However, the NF1 group appeared interested in detailed and nuanced pain tracking capabilities the expert group prioritized tracking information such as mood, nutrition, and activity to identify potential associations with pain. In tailoring the existing iCanCope application for individuals with NF1, attention should be paid to creating a community

support group feature and to tailoring content, features, and format to potential users' specific needs.

Strengths and limitations of this study

- First qualitative study to provide patient and provider perspectives about adapting a pain self-management mobile health application for individuals with NF1.
- Achieved data saturation, ensuring a deep understanding of the desired content and format when adapting the iCanCope application.
- Highlighted the need for community-based approaches as method of coping with NF1-associated chronic pain.
- The results may have limited transferability to other health conditions or mHealth applications.
- On-line focus groups limit the ability to observe in-person dynamics or subtle reactions to the various discussion topics.

INTRODUCTION

Neurofibromatosis Type 1 (NF1) is an autosomal dominant genetic disorder affecting an estimated 1 in 3000 individuals worldwide.[1] Common presentations of NF1 can include plexiform neurofibroma tumors, café au lait macules,[2, 3] and extensive dermal neurofibromas.[4] Over 50% of individuals with NF1 report significant pain and discomfort,[5] often stemming from the plexiform neurofibroma tumors.[6] In addition, frequent headaches, orthopedic, and other tumor-related pain[4] may also impair NF1 patients' quality of life.[7]

Current treatment recommendations for chronic pain related to NF1 are limited to pharmacological and surgical methods due to the complexity of the tumors.[8] It is common for individuals with NF1 to employ complementary pain management approaches (e.g., yoga, physical therapy, massage therapy) to manage their pain symptoms.[9] However, there are also challenges to receiving treatment for NF1-related chronic pain such as healthcare system barriers and limited access to self-management techniques,[10] creating a need for innovative, effective, and accessible treatment options.

Mobile health applications, with over 325,000 currently available, have become increasingly popular,[11] including some for self-management of many chronic illnesses.[12, 13] A systematic review concluded that mobile health interventions improved clinical outcomes across varying chronic diseases and increased access to pain management resources at low cost,[14] but once established, they can also be tailored to specific illnesses and conditions.

iCanCope (iCC) is an innovative mobile application originally developed to address pain self-management needs of adolescents and young adults with chronic pain.[15, 16]

The iCC platform was created through a user-centered design approach that involved end-users at every stage and is founded on the principles of cognitive behavioral therapy and the use of mind-body approaches. The core iCC features include daily symptom-tracking, personalized goal-setting to improve pain and functioning, and a toolbox of disease education and pain self-management strategies. A pilot study of iCC demonstrated feasibility of deploying the app among adolescents with chronic pain,[17] and has since been expanded to support pain self-management in youth and young adults with juvenile idiopathic arthritis,[18] sickle cell disease,[19] and post-operative pain, [20] with each adaptation reporting clinically meaningful reductions in pain intensity.

The use of mobile technology to address NF1 patients' needs is a promising strategy to expand access to pain self-management interventions. We therefore proposed to adapt the iCC platform. As a first step, we sought recommendations about the desired features and content that adults with NF1 and pain management experts considered important to include when adapting the iCC mobile health application as well as the perceived acceptability and barriers/facilitators to its use. We reasoned that the multiple perspectives of these individuals would yield a richer and more nuanced assessment of the desired features and content to include in the adaptation.

METHODS

Study Sample

The study involved two participant groups: Adults with NF1 (n = 32) and experts in the treatment and management of NF1-associated pain (n = 10). Inclusion criteria for the NF1 group were: (1) age 18-45 years with an NF1 diagnosis; (2) pain severity

aggregate score ≥3 on the Brief Pain Inventory-Short Form (BPI-SF)[21, 22] for pain experienced in the previous two weeks; (3) being proficient in English at the 5th grade level and competent to provide informed consent; (4) agreeing to audio-recording of the focus group session; and (5) residing in the U.S. The exclusion criteria consisted of: (1) moderate to severe cognitive deficits or (2) evidence of moderate to severe depression or anxiety, as assessed by the Patient Health Questionnaire (PHQ)[23] SCID-self-report (SCID-SR)[24] and Generalized Anxiety Disorder scale (GAD-7).[25] Inclusion criteria for the Expert group were: (1) providing healthcare to patients with NF1 (e.g., physicians, nurse practitioners, and psychologists) and (2) agreeing to audio-recording of the focus group session.

Patient and Public Involvement

Patients or the public were not involved in the design, or conduct, or reporting, or dissemination plans of this study.

Recruitment Procedures

Recruitment for the NF1 group consisted of posting an announcement briefly describing the study on the Facebook pages for two NF advocacy groups (NF Northeast and NF Network) and emailing all members of both group registries. Membership in both registries requires self-report of having received an NF diagnosis. Individuals interested in participating were directed to an online survey (www.qualtrics.com) that collected demographic and contact information as well as responses to the BPI, PHQ, GAD-7, and SCID-SR. The research team reviewed the results of these clinical

measures, and individuals meeting the inclusion requirements then received a list of focus group discussion (FGD) dates/times from which to select. Recruitment for the Expert group occurred via Website postings about the study on the US Association for the Study of Pain and the NF Network clinician page. Prior to beginning each FGD, participants were provided with a brief description of the study and informed of the voluntary nature of their participation. All participants provided verbal informed consent prior to participating in the FGD session.

After completing the FGD, participants in the NF1 and expert groups received a $25 and $100 gift card, respectively. The Institutional Review Board at Yale University approved all study materials and waived the need to obtain written consent.

Data Collection and Analysis

Each FGD session lasted approximately one hour and occurred via a secure, password-enforced video conference (www.zoom.com). Two members of the research team (FB, KL) with prior training in conducting focus groups jointly facilitated the sessions.

An interview guide was developed specifically for this study. Only the NF1 group sessions began by asking whether participants owned smartphones. All participants were then asked their opinions about using a phone-based application to address NF1 pain management. A brief presentation then occurred that described the existing iCC mobile application and included four screen shots from the application. The remaining session time was guided by that description, the four screen shots, and the four domains included in the FGD guide: (1) perceived barriers and facilitators to using a mobile application to manage pain; (2) perceived acceptability and benefits of the proposed application; (3) contexts within which the application would most likely be used; and (4) recommendations for possible additions or modifications to the application. Demographic information for the NF1 group included age and gender, collected at eligibility screening, and for the Expert group included professional training/title, years in practice, and gender, collected at the beginning of each FGD. The audio-recorded transcripts were professionally transcribed verbatim.

An existing codebook from a prior iCC study[26] was modified for use in the current study. Members of the data analysis team (FB, KL, LG, JS, CL) met regularly to review transcripts and discuss codebook development. Interviews continued until data saturation had been achieved.[27, 28] The final codebook included 29 codes, three of which were new codes from the Expert group sessions. Acceptable inter-coder agreement was established after independently coding two transcripts. The remaining transcripts were then independently coded by both coders (FB, KL). The coding team continued to meet periodically to resolve coding discrepancies (through consensus), address coding questions, and further refine code definitions as needed. The coded transcripts were then entered into Dedoose (www.dedoose.com, version 8.3.47) for analysis. The coding and analysis process continued in an iterative fashion using thematic analysis.[29, 30] Negative instances where the data did not fit the existing themes were identified as part of the confirmability process.[31] Descriptive statistics were calculated for the demographic data. The entire research team reviewed all analytic findings.

RESULTS

A total of nine FGDs—six NF1 groups (n = 32) and three pain management expert groups (n = 10)—occurred between January and May 2020. Previous research has shown intrinsic motivation[32] to be the critical, first step in promoting future goal-oriented behaviors.[33] We therefore sought to identify themes that shaped motivation to use the proposed mobile application and generated a total of six themes across two levels (i.e., Individual and Application) as seen in the conceptual model in Figure 1. The themes of Lifestyle, Reasons for Using the Mobile App, and Concerns regarding Use of the Mobile Application were identified at the Individual Level. The App Level included the themes of Desired Content, Desired Features, and Format Considerations.

(Insert Figure 1 here)

Description of the Study Sample

A total of 42 individuals participated in the study (Table 1). Overall, the NF1 group was generally young and predominantly female; although not formally collected, it appeared that only three participants were non-Caucasian. Except for one participant, all owned smartphones. Most within the NF1 group were experiencing moderate pain at screening. The Expert group consisted entirely of individuals with post-graduate training in their respective fields and had, on average, over 19 years of professional experience. Similar to the NF1 group, the majority of expert participants were female.

(Insert Table 1 here)

Individual Level Themes

The Individual Level focused on the life situations of individuals with NF1 that were thought to influence motivation to use the proposed mobile application.

(Insert Table 2 here)

Theme 1: Lifestyle (Individual Level)

Two sub-themes were identified within the Lifestyle theme (Table 2): Affordability/Accessibility and Time. The former focused on financial or other considerations that could influence ease of access. Both groups recognized that the mobile application needed to be “very cheap” or free and not consume much data (i.e., bottlenecking). Access issues related to geographic location or application use that depended upon external factors such as Internet signal strength or workplace access policies were also mentioned but were beyond what could be addressed within the application.

Time, the second sub-theme, focused on the busy lifestyles of most NF1 group members, usually involving parenting or work responsibilities. Both participant groups recognized that use of the application needed to easily fit into users’ schedules. Barriers to its use included forgetting to access the application during busy times or when confronted with unexpected outside demands.

Theme 2: Reasons for Using the Mobile App (Individual Level)

This theme concerned potential factors thought to directly increase motivation to use the mobile application, primarily through rapid user engagement with program content and activities. It included two sub-themes: Engagement and Hope (Table 2).

The Engagement sub-theme focused on the fact that users would be more likely to use the application if they achieved improvements or successes early on. As one expert noted, “The ultimate aim is showing progress.” Both participant groups also believed that earning rewards (e.g., badges, points) can promote engagement with and continued use of the application. Participants often referred to this as “buy-in” and viewed it as critical to program uptake.

Similarly, instilling a sense of hope was seen as essential to promoting engagement and continued use of the program. Instilling hope was not considered to be limited to explicit messages of hope but could also result from individuals with NF1 recognizing improvements in function, quality of life, or having their NF1 pain validated by others.

Theme 3: Concerns regarding Use of the Mobile App (Individual Level)

The NF1 group seemed particularly concerned about privacy and access. They worried about who would have access to their health data and whether access to such data could jeopardize health insurance coverage or pose an unforeseen risk (Table 2). As previously mentioned in the Affordability/Accessibility sub-theme, participants were also concerned that Internet coverage or work restrictions could limit use of the application. Members of the Expert group were concerned that if goals were not realistic and achievable within an appropriate timeframe, some users might discontinue using the mobile application.

The Application Level

The three themes included at the Application Level focused on elements in the proposed mobile application that could increase users' motivation to use the product. The themes primarily concerned the content to be included and the functionality and features of the proposed product.

Theme 4: Desired Content (App Level)

Participants identified the important types of information about chronic pain and its management that is needed by individuals with NF1. It included the sub-themes of "Pain 101," Self-care, and Clinical Care (Table 3).

(Insert Table 3 here)

The experts believed that accurate knowledge about the topic of pain was important in motivating individuals to use the mobile application— "a very essential piece of this work, not sufficient, but necessary" and referred to it as "Pain 101" information. This included information on the physiological basis of pain, neuropathic and nociplastic pain, the difference between acute and chronic pain, and NF1-specific pain. They also noted that users should receive instruction about how to successfully navigate the healthcare system and effectively interact with their healthcare providers.

By contrast, the NF1 group appeared most interested in learning about symptom recognition, recommended analgesics, and strategies for targeting specific types of pain.

The Self-care sub-theme dealt with pain treatment approaches potential users could learn and then do independently or in group settings. Both participant groups offered many suggestions about possible pain mitigation strategies. These could be divided into two types of self-care strategies: physical and psychobehavioral. The former, endorsed by both groups, included movement-based activities such as martial arts, yoga or “grounding exercises,” and pacing. Only the Expert group suggested improvement of sleep hygiene and use of heat and ice as physical approaches to self-management of NF1 pain. Both groups suggested psychobehavioral approaches (i.e., those requiring only brief training with a qualified professional) such as deep, circular, or meditational breathing and music or aroma therapy.

The final sub-theme, Clinical Care, focused on pain treatments that required meeting with a trained professional for longer periods than typically required in Self-care approaches. Yet, similar to the Self-care sub-theme, the treatments could be divided into physical and psychobehavioral approaches. Only the expert group identified physical approaches such as physical therapy or nutrition care. Longer term psychobehavioral approaches included cognitive behavior therapy, acceptance and commitment therapy, guided imagery, biofeedback, and mindfulness training.

Theme 5: Desired Features (App Level)

The Desired Features theme included four sub-themes: (1) Tracking capabilities, (2) Community, (3) External Resources, and (4) Future Features. This theme focused on the functionality of the mobile application and the desire for links to external resources (Table 3).

Tracking of various events and information was considered by both participant groups to be an important feature of the mobile application. It included tracking and monitoring pain level, mood, activity, sleep, and stress, all considered to be important potential pain triggers. Both groups also believed that maintaining notes or a diary was important but for somewhat different reasons. The NF1 group viewed this feature as a potential memory aid that could enable patients to “talk about [pain symptoms] more intelligently to a provider.” It could serve as a reminder for exercising, logging information in the application, or to track meditation and physical activity. The NF1 group also expressed interest in recording their surgical and medication histories and having the capability of tracking pain details such as onset, location, quality, and level of pain.

By contrast, the Expert group appeared to be interested in a less detailed approach to tracking pain, albeit sufficiently detailed to be able to identify associations between pain and such things as mood, activity, or treatment efficacy.

Both participant groups were enthusiastic about creating an online community that could serve as a resource for socioemotional support and NF1-relevant information (e.g., local events, providers). Members of the Expert group cautioned that it should not be a venue for complaining about having the disease, however. The NF1 group offered many suggestions about the format (e.g., group size, age restrictions, creation of a buddy system), although none emerged as the preferred format. Both groups also agreed that sharing experiences could help normalize having NF1 and help individuals feel less isolated.

Participants believed that external resources such as links to other mobile applications could be helpful to NF1 patients. The NF1 group was interested in links to relevant articles and treatment guidelines. The Expert group suggested links that could assist individuals with NF1 in self-management of their disease, including relaxation and pain sites.

Both groups identified some features that were currently not feasible to include in the mobile application, as they would require extensive resources, negotiations with other applications, or lengthy modifications to the current iCC. They can be considered for future modifications to the application. Suggestions included automatic data upload at the next sign-in to make it more convenient for users whose Internet access is blocked at work or in certain geographical locations, printable reports, 24/7 access to medical advice, and a visual graphic representation of progress toward a specific goal.

Other suggestions, primarily from the NF1 group, were currently unrealistic to implement because they require formalizing agreements with healthcare institutions. For example, they mentioned wanting the capability to automatically share information with healthcare providers or access existing x-rays when at another location.

Theme 6: Format Considerations (App Level)

The final theme included two sub-themes (Table 3): User Interface and Presentation/Tone. The first addressed issues that would increase users' ease of use, and the second focused on the visual appeal and tone of the content presented.

Both groups understood the importance of User Interface, that motivation to use the mobile application could be heavily influenced by its ease of use. The Expert group suggested having explicit instructions on how to navigate and use the application. Participants from both groups recommended tailoring the application to users' specific preferences on pain-tracking measures and format (e.g., numeric vs. figural, pain descriptors) and including an "Other" option so users could further qualify their responses to forced-choice questions. They also identified features to enhance time-efficiency such as auto-population of key information, use of drop-down menus, and consistency in format and content when updating.

Critical elements in motivating individuals to use the mobile application were Tone and Presentation, the final sub-theme. Participants described the ideal application as being "approachable and digestible" and "simple and fun." Visual appeal was important, noting that the font and use of color could influence use. Careful selection of NF1 photographs and language could also be used to set an appropriate tone. Members of the NF1 group believed that daily tips or inspiring quotes could enhance engagement, and experts recommended varying the messages to keep the content fresh. Both groups agreed that music could help set a positive tone as well.

DISCUSSION

Few studies have examined the perceived benefits, desired content and features, or efficacy of mobile health applications on coping with NF1-related chronic pain. To our knowledge, the current study is the only one to examine some of these issues from the perspectives of the targeted end-users and pain management experts. We believe that this is essential information to ensure that the product is scientifically accurate and relevant while also satisfying targeted end-users' needs and preferences.

Both groups were enthusiastic about the prospect of adapting the iCC mobile application for individuals with NF1 with chronic pain. They believed that tailoring the program to accommodate users' lifestyle needs and affordability, incentivizing its use, fostering an NF1 community support group, and using appropriate and acceptable tone and format features will encourage uptake and continued use of the mobile application. In addition, offering NF1 and coping skills information and an online support community in a single mobile application renders it both convenient and comprehensive in scope.

Participants believed that flexibility in fitting into potential users' current situation was important. They noted that the application should be affordable, easy to use, and convenient; it should fit users' schedules, efficiently capture the desired data, and be modifiable as users' life situations change. These findings support previous research on features needed to ensure uptake and ease of use of the mobile application[34, 35] as well as health-related mobile application users' privacy and security concerns.[36, 37]

With respect to the issue of promoting sustained use of the application, participants from both groups noted the importance of including realistic and achievable goals. Early improvements might involve pain reduction or changes in pain attitudes that could improve mood, function, or quality of life. Consistent with the notion of contingency management,[38, 39] participants identified strategies involving points, levels, prizes, or well-established reinforcement schedules to incentivize continued use. These findings suggest the importance of considering contingency management when developing an application to promote health behaviors.[40,41]

Regarding the pain symptom tracking feature, the two groups had somewhat different views on what type of pain data are most desired. The NF1 group appeared interested in gathering as detailed and nuanced pain information as possible. By contrast, the Expert group appeared more interested in tracking information that might identify potential associations between pain, mood, nutrition, activity, and stress levels; they believed that noting these patterns would be more useful than tracking pain intensity, duration, quality, and directionality. Consistent with previous research,[42] the two participant groups expressed different perspectives on and desires for tracking chronic pain. The data suggest that to accommodate these disparate views, the pain tracking feature should permit simultaneous tracking of several constructs (e.g., pain, mood, activity) while also providing a flexible format so that patients can select the level of detail and pain metrics they prefer. One cautionary note, however, would be to avoid the potential for users to become fixated on their pain symptoms to the detriment of recognizing the associations between their pain and the other constructs.

Community support, particularly for individuals with rare diseases such as NF1, can be an important coping resource.[43,44] Previous research on online community support platforms for individuals with rare diseases has noted that they are used for both informational and socioemotional support purposes.[45, 46] The value of community support has been identified within the context of cultural and racial socialization,[47, 48] and the study corroborates these findings. Both groups noted how the creation of an online NF1 community would provide users the opportunity to share and thereby validate their experiences with the disease and chronic pain, provide socioemotional encouragement, and potentially motivate continued use of the mobile application.

The study supported previous findings about visual appeal, tone, ease of use, instructions on effectively navigating app options, tailoring mHealth interventions to personal needs and preferences, and the use of appropriate visuals.[49, 50] This last feature is particularly important with a potentially disfiguring disease such as NF1. A novel finding from this study, however, is the importance of establishing an upbeat tone that instills hope.

We note several study limitations. First, the findings may have limited transferability but we instead sought multiple perspectives on potential issues to consider when adapting the iCC for use by those with NF1. Second, the myriad suggestions proffered may be attributed to social desirability wherein participants sought to identify as many options as possible in order to "please" the group facilitators. Nonetheless, these options made possible a deeper analysis of potential format and content to be considered when adapting the iCC application. Finally, virtual FGDs limit the ability to observe subtle, in-person dynamics during discussions. We chose this format, however, prior to the onset of the COVID-19 pandemic, based upon our desire to collect data in a time- and cost-efficient manner across diverse geographic regions and living situations.

Conclusions

The study findings suggest that adapting the iCC mobile application for use in the self-management of NF1-associated chronic pain was seen by adults with NF1 and chronic pain experts as a welcomed addition to the armamentarium of NF1 treatment resources. Careful attention should be paid to the creation of a community support group option as well as program content, format, and ability to tailor it to users' specific needs.

Acknowledgements: We are indebted to Karen Peluso, Director of NF Northeast and Kim Bischoff, Director of NF Network for their help and commitment. We also thank the study participants for sharing with us their time and insights.

Funding: This work was supported by the U.S. Army Medical Research Materiel Command endorsed by the U.S. Army, through the Congressionally Directed Medical Research Programs' Neurofibromatosis Research Program under Award No. W81XWH-19-1-0618.

Competing Interests Statement: None declared.

Author Contributions:

LG- Manuscript preparation and review, study design and data analysis, development of interview guide

KL- Daily oversight of project, focus group facilitation, data management and analysis, manuscript preparation and review

CL- Development of interview guide, manuscript preparation and review

JS- Development of interview guide, manuscript preparation and review

WZ- Supervision of the project, manuscript review

SB- Supervision of the project, manuscript review

FB- Study design and data analysis, led the focus groups, development of interview guide, manuscript preparation and review, oversight for the general conduct of this study

Data Sharing Statement: Requests to receive the qualitative data should be addressed to the Corresponding Author. Upon review of the request, the data may be made available.

Ethics Statement: The Human Subjects Committee of the Yale Human Research Protection Program of Institutional Review Boards at Yale University approved Protocol #2000025437.

REFERENCES

1. Anderson JL, Gutmann DH. Neurofibromatosis type 1. *Handb Clin Neurol* 2015;132:75-86. PMID: 26564071. doi: 10.1016/B978-0-444-62702-5.00004-4.
2. Kodra Y, Giustini S, Divona L, Porciello R, Calvieri S, Wolkenstein P, et al. Health-related quality of life in patients with neurofibromatosis type 1. A survey of 129 Italian patients. *Dermatology* 2009;218:215-20. PMID: 19088462. doi: 10.1159/000187594.
3. Radtke HB, Sebold CD, Allison C, Haidle JL, Schneider G. Neurofibromatosis type 1 in genetic counseling practice: recommendations of the National Society of Genetic Counselors. *J Genet Couns* 2007;16:387-407. PMID: 17636453. doi: 10.1007/s10897-007-9101-8.
4. Tonsgard JH. Clinical manifestations and management of neurofibromatosis type 1. *Semin Pediatr Neurol* 2006;13:2-7. PMID: 16818170. doi: 10.1016/j.spen.2006.01.005.
5. Brems H, Beert E, de Ravel T, Legius E. Mechanisms in the pathogenesis of malignant tumours in neurofibromatosis type 1. *Lancet Oncol* 2009;10:508-15. PMID: 19410195. doi: 10.1016/s1470-2045(09)70033-6.
6. North KN, Riccardi V, Samango-Sprouse C, Ferner R, Moore B, Legius E, et al. Cognitive function and academic performance in neurofibromatosis. 1: consensus statement from the NF1 Cognitive Disorders Task Force. *Neurology* 1997;48:1121-7. PMID: 9109916. doi: 10.1212/wnl.48.4.1121.
7. Vranceanu A-M, Merker VL, Park E, Plotkin SR. Quality of life among adult patients with neurofibromatosis 1, neurofibromatosis 2 and schwannomatosis: a systematic review of the literature. *J Neurooncol* 2013;114:257-62. doi: 10.1007/s11060-013-1195-2.
8. Sabatini C, Milani D, Menni F, Tadini G, Esposito S. Treatment of neurofibromatosis type 1. *Curr Treat Options Neurol* 2015;17(6):355. PMID: 25917340. doi: 10.1007/s11940-015-0355-4.
9. Buono FD, Grau LE, Sprong ME, Morford KL, Johnson KJ, Gutmann DH. Pain symptomology, functional impact, and treatment of people with Neurofibromatosis type 1. J *Pain Res* 2019;12:2555-61. PMID: 31692483. doi: 10.2147/jpr.S209540.
10. Martin S, Wolters PL, Toledo-Tamula MA, Schmitt SN, Baldwin A, Starosta A, et al. Acceptance and commitment therapy in youth with neurofibromatosis type 1 (NF1) and chronic pain and their parents: A pilot study of feasibility and preliminary efficacy. *Am J Med Genet A* 2016;170:1462-70. PMID: 27021207. doi: 10.1002/ajmg.a.37623.
11. Ferrara G, Kim J, Lin S, Hua J, Seto E. A Focused Review of Smartphone Diet-Tracking Apps: Usability, Functionality, Coherence With Behavior Change Theory, and Comparative Validity of Nutrient Intake and Energy Estimates. *JMIR Mhealth Uhealth* 2019;7:e9232. PMID: 31102369. doi: 10.2196/mhealth.9232.
12. Birkhoff SD, Smeltzer SC. Perceptions of Smartphone User-Centered Mobile Health Tracking Apps Across Various Chronic Illness Populations: An Integrative

Review. *J Nurs Scholarsh* 2017;49:371-8. PMID: 28605151. doi: 10.1111/jnu.12298.

13. Sarkar U, Gourley GI, Lyles CR, Tieu L, Clarity C, Newmark L, et al. Usability of Commercially Available Mobile Applications for Diverse Patients. *J Gen Intern Med* 2016;31:1417-26. PMID: 27418347. doi: 10.1007/s11606-016-3771-6.
14. Whitehead L, Seaton P. The Effectiveness of Self-Management Mobile Phone and Tablet Apps in Long-term Condition Management: A Systematic Review. *J Med Internet Res* 2016;18:e97. PMID: 27185295. doi: 10.2196/jmir.4883.
15. Stinson JN, Lalloo C, Harris L, Isaac L, Campbell F, Brown S, et al. iCanCope with Pain™: User-centred design of a web- and mobile-based self-management program for youth with chronic pain based on identified health care needs. *Pain Res Manag* 2014;19:257-65. PMID: 25000507. doi: 10.1155/2014/935278.
16. Stinson JN, White M, Isaac L, Campbell F, Brown S, Ruskin D, et al. Understanding the information and service needs of young adults with chronic pain: perspectives of young adults and their providers. *Clin J Pain* 2013;29:600-12. PMID: 23328333. doi: 10.1097/AJP.0b013e31826dce65.
17. Lalloo C, Hundert A, Harris L, Pham Q, Campbell F, Chorney J, et al. Capturing Daily Disease Experiences of Adolescents With Chronic Pain: mHealth-Mediated Symptom Tracking. *JMIR Mhealth Uhealth* 2019;7:e11838. PMID: 30664472. doi: 10.2196/11838.
18. Lalloo C, Harris LR, Hundert AS, Berard R, Cafazzo J, Connelly M, et al. The iCanCope pain self-management application for adolescents with juvenile idiopathic arthritis: a pilot randomized controlled trial. *Rheumatology (Oxford)* 2021;60:196-206. PMID: 32613229. doi: 10.1093/rheumatology/keaa178.
19. Palermo TM, Zempsky WT, Dampier CD, Lalloo C, Hundert AS, Murphy LK, et al. iCanCope with Sickle Cell Pain: Design of a randomized controlled trial of a smartphone and web-based pain self-management program for youth with sickle cell disease. *Contemp Clin Trials* 2018;74:88-96. PMID: 30316999. doi: 10.1016/j.cct.2018.10.006.
20. Birnie KA, Campbell F, Nguyen C, Lalloo C, Tsimicalis A, Matava C, et al. iCanCope PostOp: User-Centered Design of a Smartphone-Based App for Self-Management of Postoperative Pain in Children and Adolescents. *JMIR Form Res* 2019;3:e12028. PMID: 31008704. doi: 10.2196/12028.
21. Cleeland CS, Ryan KM. Pain assessment: global use of the Brief Pain Inventory. *Ann Acad Med Singap* 1994;23:129-38. PMID: 8080219.
22. Palos GR, Mendoza TR, Mobley GM, Cantor SB, Cleeland CS. Asking the community about cutpoints used to describe mild, moderate, and severe pain. *J Pain* 2006;7:49-56.
23. Kroenke K, Spitzer RL, Williams JB. The PHQ-9: validity of a brief depression severity measure. *J Gen Intern Med* 2001;16:606-13. PMID: 11556941. doi: 10.1046/j.1525-1497.2001.016009606.x.

24. Brodey B, Purcell SE, Rhea K, Maier P, First M, Zweede L, et al. Rapid and Accurate Behavioral Health Diagnostic Screening: Initial Validation Study of a Web-Based, Self-Report Tool (the SAGE-SR). *J Med Internet Res* 2018;20:e108. PMID: 29572204. doi: 10.2196/jmir.9428.

25. Spitzer RL, Kroenke K, Williams JB, Lowe B. A brief measure for assessing generalized anxiety disorder: the GAD-7. *Arch Intern Med* 2006;166):1092-7. PMID: 16717171. doi: 10.1001/archinte.166.10.1092.

26. Ahola Kohut S, LeBlanc C, O'Leary K, McPherson AC, McCarthy E, Nguyen C, et al. The internet as a source of support for youth with chronic conditions: A qualitative study. *Child Care Health Dev* 2018;44:212-20. PMID: 29082537. doi: 10.1111/cch.12535.

27. Guest G, Bunce A, Johnson L. How Many Interviews Are Enough?:An Experiment with Data Saturation and Variability. *Field Methods* 2006;18:59-82. doi: 10.1177/1525822x05279903.

28. Hennink MM, Kaiser BN, Marconi VC. Code Saturation Versus Meaning Saturation: How Many Interviews Are Enough? *Qual Health Res* 2017;27:591-608. PMID: 27670770. doi: 10.1177/1049732316665344.

29. Braun V, Clarke V. Using thematic analysis in psychology. Qualitative Research in Psychology. 2006 2006/01/01;3(2):77-101. doi: 10.1191/1478088706qp063oa.

30. Braun V, Clarke V. Successful qualitative research: A practical guide for beginners: sage; 2013. ISBN: 1446289516.

31. Henwood KL, Pidgeon NF. Qualitative research and psychological theorizing. Br J Psychol. 1992 Feb;83 (Pt 1):97-111. PMID: 1559146. doi: 10.1111/j.2044-8295.1992.tb02426.x.

32. Deci EL, Ryan RM. Intrinsic Motivation and Self-Determination in Human Behavior.1985. New York:Springer Science+Business Media. ISBN: 978-1-4899-2273-1.

33. Ajzen I. From Intentions to Actions: A Theory of Planned Behavior. In: Kuhl J, Beckmann J, editors. Action Control: From Cognition to Behavior. Berlin:Springer-Verlag Berlin Heidelberg; 1985. p. 11-39. ISBN: 978-3-642-69746-3

34. O'Brien T, Rosenthal A. Preferred Features in Mobile Health Applications for Kidney Transplant Recipients: A Qualitative Approach. *Nephrol Nurs J* 2020;47:529-36. [published Online First: 2020/12/31]

35. Zapata BC, Fernández-Alemán JL, Idri A, et al. Empirical Studies on Usability of mHealth Apps: A Systematic Literature Review. *J Med Syst* 2015;39:1. doi: 10.1007/s10916-014-0182-2

36. Martínez-Pérez B, de la Torre-Díez I, López-Coronado M. Privacy and security in mobile health apps: a review and recommendations. *J Med Syst* 2015;39:181. doi: 10.1007/s10916-014-0181-3

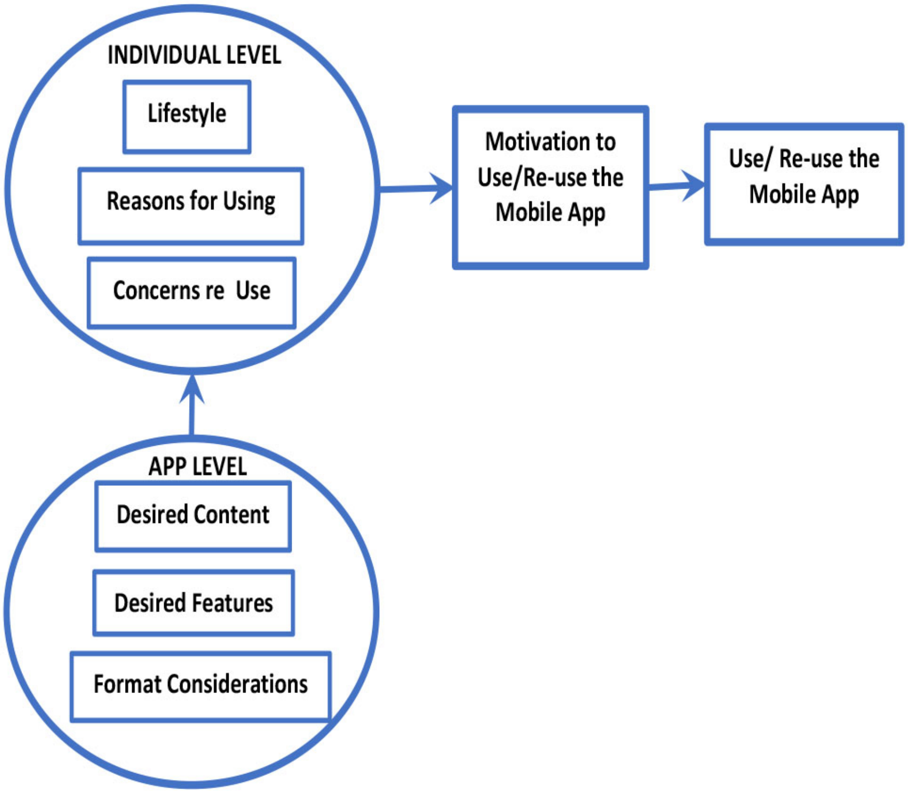
37. Agarwal P, Gordon D, Griffith J, Kithulegoda N, Witteman HO, Sacha Bhatia R, et al. Assessing the quality of mobile applications in chronic disease management: a

scoping review. *NPJ Digit Med* 2021;4:46. PMID: 33692488. doi: 10.1038/s41746-021-00410-x.

38. Higgins ST, Petry NM. Contingency management. Incentives for sobriety. *Alcohol Res Health* 1999;23:122-7. PMID: 10890806.
39. Higgins ST, Silverman K. Motivating behavior change among illicit-drug abusers: Research on contingency management interventions. Washington, DC, US: American Psychological Association; 1999. ISBN: 1-55798-570-7.
40. Máca V, Ščasný M, Zvěřinová I, et al. Incentivizing Commuter Cycling by Financial and Non-Financial Rewards. *Int J Environ Res Public Health* 2020;17:6033 doi: 10.3390/ijerph17176033
41. Carpenter VL, Hertzberg JS, Kirby AC, et al. Multicomponent smoking cessation treatment including mobile contingency management in homeless veterans. *J Clin Psychiatry* 2015;76:959-64. doi: 10.4088/JCP.14m09053
42. Kovačević I, Kogler VM, Turković TM, Dunkić LF, Ivanec Ž, Petek D. Self-care of chronic musculoskeletal pain - experiences and attitudes of patients and health care providers. *BMC Musculoskelet Disord* 2018;19:76. PMID: 29514616. doi: 10.1186/s12891-018-1997-7.
43. Martin S, Struemph KL, Poblete A, et al. An Internet support group for parents of children with neurofibromatosis type 1: a qualitative analysis. *J Community Genet* 2018;9:327-34. doi: 10.1007/s12687-018-0360-x
44. Martin S, Wolters PL, Baldwin A, et al. Attitudes about internet support groups among adolescents and young adults with neurofibromatosis type 1 and their parents. *J Genet Couns* 2014;23:796-804. doi: 10.1007/s10897-014-9688-5
45. Lasker JN, Sogolow ED, Sharim RR. The role of an online community for people with a rare disease: content analysis of messages posted on a primary biliary cirrhosis mailinglist. *J Med Internet Res* 2005;7:e10. PMID: 15829472. doi: 10.2196/jmir.7.1.e10.
46. Doyle M. Peer Support and Mentorship in a US Rare Disease Community: Findings from the Cystinosis in Emerging Adulthood Study. *Patient* 2015;8:65-73. PMID: 25231828. doi: 10.1007/s40271-014-0085-9.
47. Litchman ML, Edelman LS, Donaldson GW. Effect of Diabetes Online Community Engagement on Health Indicators: Cross-Sectional Study. *JMIR Diabetes* 2018;3:e8. doi: 10.2196/diabetes.8603
48. Young SD, Holloway I, Jaganath D, et al. Project HOPE: online social network changes in an HIV prevention randomized controlled trial for African American and Latino men who have sex with men. *Am J Public Health* 2014;104:1707-12. doi: 10.2105/ajph.2014.301992
49. Llorens-Vernet P, Miró J. Standards for Mobile Health-Related Apps: Systematic Review and Development of a Guide. *JMIR Mhealth Uhealth* 2020;8:e13057. PMID: 32130169. doi: 10.2196/13057.
50. Woods L, Duff J, Cummings E, Walker K. Evaluating the Development Processes of Consumer mHealth Interventions for Chronic Condition Self-management: A

Scoping Review. *Comput Inform Nurs* 2019;37:373-85. PMID: 31135468. doi: 10.1097/cin.0000000000000528.

Figure 1 Conceptual Framework and Themes for Motivation to Use the Mobile Application


INDIVIDUAL LEVEL
Lifestyle
Reasons for Using
Concerns re Use
Motivation to Use/Re-use the Mobile App
Use/ Re-use the Mobile App
APP LEVEL
Desired Content
Desired Features
Format Considerations

Six themes identified within the data were thought to shape motivation to use the mobile application. Three could be attributed to personal characteristics of the targeted end-users and were thought to directly influence motivation. The remaining three focused on content and qualities of the application itself and were thought to indirectly influence motivation to use the application.

90x90mm (300 x 300 DPI)

Table 1. Characteristics of the Study Sample (N = 42)

	NF1 Group (n = 32)	Expert Group (n = 10)
Age (years)[1]	31.2 (4.9)	—
Range	18 - 39	
Gender[2]		
Male	6 (19%)	4 (40%)
Female	25 (78%)	6 (60%)
Other	1 (4%)	—
Aggregate Pain Severity[1,3]	4.8 (1.7)	—
Aggregate Pain Interference[1,4]	4.9 (2.6)	—
Professional Training/Title[2]		
Pain Psychologist	—	2 (20%)
Neurologist	—	5 (50%)
Primary Care Physician	—	2 (20%)
Neuro-oncologist	—	1 (10%)
Years in Practice[1]	—	19.2 (7.2)
Range		7 - 38

[1] Mean (S.D.)
[2] N (%)
[3] BPI-SF; 4 items for pain severity, possible range of scores is 0 - 10
[4] BPI-SF, 7 items for pain interference, possible range of scores is 0 - 10

Table 2. Individual Level Themes and Quotes

Theme 1: Lifestyle	
Affordability/Accessibility sub-theme	Where I work there's certain applications that are blocked so it would be really a pain for lack of a better term for me to have to kind of remember everything and write it down later. (Participant 26, NF1 group)
	Unfortunately, in rural areas such as southwest Pennsylvania where I live or West Virginia a lot of people just don't have connections that are that strong sometimes. (Participant 24, NF1 group)
	There's always a lot of barriers in terms of access, economics, et cetera. (Participant 103, Expert group)
Time sub-theme	Sometimes I get up late and it's just something that I need to do every day but tend to forget if I'm hustling out the door or something. (Participant 7, NF1 group)
	I'm very bad about inputting stuff and keeping diaries and stuff because I have two toddlers at home, so that takes a lot of my time, too. (Participant 17, NF1 group)
	Other barriers are typically time… (Participant 106, Expert group)
Theme 2: Reasons for Using the Mobile Application	
Engagement sub-theme	I like apps that give me badges and awards for accomplishing my goals. (Participant 10, NF1 group)
	There has to be a way that they can, early in their treatment, start to see positive change, positive success so they get these little wins in the beginning and that's just the psychology of motivation and how you sort of really start to get that ball rolling. (Participant 104, Expert group)
Hope sub-theme	They have to be able to hold hope that they have the opportunity to be efficacious in improving the situation. (Participant 104, Expert group)
Theme 3: Concerns about Using the App	Who could potentially access that information? Could health insurance companies access…I'm worried about who has access to that and how they could potentially use it. (Participant 14, NF1 group)
	[Use of stars or points] can be very effective if they're tooled in the right way…but they [users] have to feel like that the goals are attainable. (Participant 104, female, Expert group)

Table 3. Application Level Themes and Quotes

Theme 4: Desired Content	
"Pain 101" sub-theme	There's an abundance of research also on pain education as a pain management strategy…I do think the pain education piece and how pain works in the brain would be really great for NF patients. (Participant 101, Expert group)
	…how to work through and navigate the healthcare system, how to talk to healthcare providers about their pain and options. (Participant 106, Expert group)
	It's the specific exercises or to target or…to alleviate that specific pain. To have it be specific, broken down like a menu. Headaches? Do this. Sciatic pain? Do that. (Participant 2, NF1 group)
Self-care sub-theme	I think the mindfulness or meditation, yoga, breathing techniques would be extremely helpful. (Participant 26, NF1 group)
	…just getting them exposed to different complementary and alternative therapies has value, so even if you don't teach it in the app, if there was even a little module that was just like things you could try. (Participant 108, Expert group)
Clinical Care sub-theme	I did a study for NF with ACT, that's Acceptance and Commitment Treatment. I liked it because I think if you were going to do something with CBT [cognitive-behavior therapy], if you could incorporate some ACT as well, that would be great because to help you rethink about your pain might be, for me at least, it's helped me kind of restructure myself a little bit. (Participant 10, NF1 group)
	Cognitive behavioral therapy, evidence-based treatment for chronic pain involves a whole host of cognitive and behavioral strategies including pacing for pain and mindfulness, relaxation, catching catastrophic thoughts about pain and about the future, as well as lifestyle changes like nutrition and sleep hygiene. (Participant 101, Expert group)
Theme 5: Desired Features	
Tracking Capabilities sub-theme	I had to go through boxes of medical records to figure out and remember all the different doctors…so if there was a way to store some information… (Participant 3, NF1 group)
	I think one cool thing would be if there was an outline of a body that you could blow up and click on the part exactly that hurts and kind of color it in or shade it in or something" (Participant 19, NF1 group)
	I really struggle in striking the balance between having data on the pain but then also being able to do something about it…It's one thing if a patient came in and said, "Here's the graph, here's how much pain I've been having." I can look at that say, "Oh, that's rough. What are we going to do about that?" But if here's a graph then he or she also says here's the pain I was having and here's what else what was going on at that time and what I think may have contributed, then I may be able to offer some further intervention. (Participant 109, Expert group)
	I think tracking mood is hugely important. We know that anxiety and depression very commonly and understandably co-occur with chronic pain and medical conditions. We also know that mood and stress and anxiety and depression impact and amplify pain, so I definitely think it's important to assess mood and also to explain that mood and emotion is directly linked to the experience of pain. (Participant 101, Expert group)
Community sub-theme	…things where we could actually keep up with one another, talk to one another, just get things off our chest. I think that would be incredibly beneficial. (Participant 6, NF1 group)
	I think having positive or some opportunities to connect with other people that really understand the disease entity can be helpful too, as long as they're all working toward making gains and not just being really steeped and sad about the disease process. (Participant 104, Expert group)

	It was years before I ever met anybody else with NF and it's nice to know that there are other people out there…it would be nice on this app to be able to have maybe a comment section so that people can say, yup, I was there, or this is how I cope. (Participant 14, NF1 group)
	…people share incredible stuff on [message boards], so sharing pain may be nothing compared to sharing their personal stories about all their tumors and talking about a doctor. (Participant 107, Expert group)
External Resources sub-theme	I think of it as a way to maybe link certain maybe research articles or advice [that]… afterwards if you log a specific instance of pain and there's something that could pop up that you could read or if you log something that's maybe abnormal based on what you've logged in the past it could provide you some kind of feedback. (Participant 6, NF1 group)
	We use [name of app] …And it's been really helpful for things like CRPS [complex regional pain syndrome], potentially amputation pain, and so it's part of cognitive retraining for the disorganized cognitive input from a missing limb or a dysfunctional limb. (Participant 105, Expert group)
Future Features sub-theme	One thing I'd like to see for the physical activity, if it could import data from say [names of activity apps] or whatever if you use one of those apps, it would be one less thing for the user to do. (Participant 20, NF1 group)
	I'm a software developer myself so I want my app to be able to function at least as good as it can with no Internet connection and automatically update the database when I get connection. (Participant 24, NF1 group)
	One of the apps I used to use was for water and as you drank water it grew a plant. Well, maybe something interactive based on the age ranges of who might use it. (Participant 9, NF1 group)
	…if [patients] can print out a graph or somehow send the graphical representation that we've like scanned that into the medical record, that's been helpful (Participant 108, Expert group)
	I think one really important feature that a lot of other applications that I use lack is integration with MyChart and other things that my doctors would have access to so that everybody can just see it rather than having to go back and forth with all the different doctors (Participant 26, NF1 group)
Theme 6: Format Considerations	
User Interface sub-theme	I need consistency. Kind of like if you're going to do updates and stuff like that, it needs to not be completely reconfigured. (Participant 1, NF1 group)
	If I can add in my own entries along with that, that would motivate me to use it more. (Participant 18, NF1 group)
	…then you want a very simple front page and then links or tabs. (Participant 105, Expert group)
Presentation/Tone sub-theme	I think having some visuals as well would be useful. Yeah, visual, not horrid looking. (Participant 1, NF1 group)
	I would want it to be simple and fun and use language that I use, not language a doctor would use. I can look up words, but sometimes I don't want to. (Participant 4, NF1 group)
	I think considering color is important…We didn't really talk about that but, the whole visual appeal. Because if it's not appealing or if it's going to make a person feel worse then that's counter, then that's going against what [the goal of the application is]. (Participant 102, Expert group)
	When I was looking at your app, it made me think about music and how that can be useful for people's state of mind, which you can't separate the body from the mind so I think that would be great in chronic pain. (Participant 103, Expert group)

BMJ Open

Perspectives on Adapting a Mobile Application for Pain Self-Management in Neurofibromatosis Type 1: Results of Online Focus Group Discussions with Individuals Living with Neurofibromatosis Type 1 and Pain Management Experts

Journal:	*BMJ Open*
Manuscript ID	bmjopen-2021-056692.R1
Article Type:	Original research
Date Submitted by the Author:	04-Apr-2022
Complete List of Authors:	Grau, Lauretta; Yale University Yale School of Public Health, Department of Epidemiology of Microbial Diseases Larkin, Kaitlyn; Yale School of Medicine, Department of Psychiatry Lalloo, Chitra; The Hospital for Sick Children, Child Health Evaluative Sciences; University of Toronto, Institute of Health Policy, Management & Evaluation Stinson, Jennifer; Toronto SickKids, Child Health Evaluative Sciences; University of Toronto, Institute of Health Policy, Management & Evaluation Zempsky, William; University of Connecticut School of Medicine, Department of Pediatrics and Nursing; Connecticut Children's Medical Center, Division of Pain and Palliative Care Ball, Samuel; Yale School of Medicine, Department of Psychiatry Buono, Frank; Yale School of Medicine, Department of Psychiatry
Primary Subject Heading:	Complementary medicine
Secondary Subject Heading:	Neurology
Keywords:	COMPLEMENTARY MEDICINE, Adult neurology < NEUROLOGY, Neurogenetics < NEUROLOGY, PAIN MANAGEMENT, QUALITATIVE RESEARCH, Neurological pain < NEUROLOGY

SCHOLARONE™
Manuscripts

I, the Submitting Author has the right to grant and does grant on behalf of all authors of the Work (as defined in the below author licence), an exclusive licence and/or a non-exclusive licence for contributions from authors who are: i) UK Crown employees; ii) where BMJ has agreed a CC-BY licence shall apply, and/or iii) in accordance with the terms applicable for US Federal Government officers or employees acting as part of their official duties; on a worldwide, perpetual, irrevocable, royalty-free basis to BMJ Publishing Group Ltd ("BMJ") its licensees and where the relevant Journal is co-owned by BMJ to the co-owners of the Journal, to publish the Work in this journal and any other BMJ products and to exploit all rights, as set out in our licence.

The Submitting Author accepts and understands that any supply made under these terms is made by BMJ to the Submitting Author unless you are acting as an employee on behalf of your employer or a postgraduate student of an affiliated institution which is paying any applicable article publishing charge ("APC") for Open Access articles. Where the Submitting Author wishes to make the Work available on an Open Access basis (and intends to pay the relevant APC), the terms of reuse of such Open Access shall be governed by a Creative Commons licence – details of these licences and which Creative Commons licence will apply to this Work are set out in our licence referred to above.

Other than as permitted in any relevant BMJ Author's Self Archiving Policies, I confirm this Work has not been accepted for publication elsewhere, is not being considered for publication elsewhere and does not duplicate material already published. I confirm all authors consent to publication of this Work and authorise the granting of this licence.

Perspectives on Adapting a Mobile Application for Pain Self-Management in Neurofibromatosis Type 1: Results of Online Focus Group Discussions with Individuals Living with Neurofibromatosis Type 1 and Pain Management Experts

Lauretta E. Grau, PhD[1], Kaitlyn Larkin, BA[2], Chitra Lalloo, PhD[3,4], Jennifer N. Stinson, RN, PhD[3,4], William T. Zempsky, MD[5,6], Samuel A. Ball PhD[2], and Frank D. Buono, PhD[2]

[1] Department of Epidemiology of Microbial Diseases, Yale School of Public Health, New Haven, CT USA
[2] Department of Psychiatry, Yale School of Medicine, New Haven, CT USA
[3] Child Health Evaluative Sciences, The Hospital for Sick Children, Toronto, Ontario
[4] Institute of Health Policy, Management & Evaluation, University of Toronto, Toronto, Ontario
[5] Department of Pediatrics and Nursing, University of Connecticut, Hartford, CT, USA
[6] Division of Pain and Palliative Medicine, Connecticut Children's Medical Center, Hartford, CT, USA

Corresponding Author:
Lauretta E. Grau, PhD
Yale School of Public Health
PO Box 208034
New Haven, CT 06520
E: lauretta.grau@yale.edu

Key words: COMPLEMENTARY MEDICINE, Adult Neurology, Neurological pain, Neurogenetics, PAIN MANAGEMENT, QUALITATIVE RESEARCH

Word count: 3873; abstract 272

ABSTRACT

Objectives: Neurofibromatosis Type 1 (NF1) is a genetic disorder in which chronic pain commonly occurs. The study sought to understand the needs of individuals with NF1 and pain management experts when adapting a pain self-management mobile health application for individuals with NF1. **Design:** We conducted a series of online, audio-recorded focus groups that were then thematically analyzed. **Setting:** Online focus groups with adults with NF1 currently residing in the U.S. **Participants:** Two types of participants were included: Individuals with NF1 (n = 32 across six focus groups) and pain management experts (n = 10 across three focus groups). **Results:** Six themes across two levels were identified. The Individual level included Lifestyle, Reasons for Using the Mobile App, and Concerns regarding its Use. The Application level included Desired Content, Desired Features, and Format Considerations. Findings included recommendations to grant free access to the application and include a community support feature for individuals to relate and validate one another's experience with pain from NF1. In addition, participants noted the importance of providing clear instructions on navigating the app, the use of an upbeat, hopeful tone, and appropriate visuals.

Conclusions: Both participant groups endorsed the use of iCanCope as an NF1 pain self-management mobile application. Differences between groups was noted regarding However, the NF1 group appeared interested in detailed and nuanced pain tracking capabilities the expert group prioritized tracking information such as mood, nutrition, and activity to identify potential associations with pain. In tailoring the existing iCanCope application for individuals with NF1, attention should be paid to creating a community

support group feature and to tailoring content, features, and format to potential users' specific needs.

Strengths and limitations of this study

- First qualitative study to include both patient and provider perspectives about adapting a pain self-management mobile health application for individuals with NF1.
- Achieved data saturation, ensuring a deep understanding of the stated research question.
- Online focus group format permitted a time- and cost-efficient opportunity to enable wide geographic representation among participants.
- Unverified NF1 status and low representation among members of minority communities may have limited the credibility and transferability of the findings.
- On-line focus groups limit the ability to observe in-person dynamics or subtle reactions to the various discussion topics.

INTRODUCTION

Neurofibromatosis Type 1 (NF1) is an autosomal dominant genetic disorder affecting an estimated 1 in 3000 individuals worldwide.[1] Common presentations of NF1 can include plexiform neurofibroma tumors, café au lait macules,[2, 3] and extensive dermal neurofibromas.[4] Over 50% of individuals with NF1 report significant pain and discomfort,[5] often stemming from the plexiform neurofibroma tumors.[6] In addition, frequent headaches, orthopedic, and other tumor-related pain[4] may also impair NF1 patients' quality of life.[7]

Current treatment recommendations for chronic pain related to NF1 are limited to pharmacological and surgical methods due to the complexity of the tumors.[8] It is common for individuals with NF1 to employ complementary pain management approaches (e.g., yoga, physical therapy, massage therapy) to manage their pain symptoms.[9] However, there are also challenges to receiving treatment for NF1-related chronic pain such as healthcare system barriers and limited access to self-management techniques,[10] creating a need for innovative, effective, and accessible treatment options.

Mobile health applications, with over 325,000 currently available, have become increasingly popular,[11] including some for self-management of many chronic illnesses.[12, 13] A systematic review concluded that mobile health interventions improved clinical outcomes across varying chronic diseases and increased access to pain management resources at low cost,[14] but once established, they can also be tailored to specific illnesses and conditions.

iCanCope (iCC) is an innovative mobile application originally developed to address pain self-management needs of adolescents and young adults with chronic pain.[15, 16]

The iCC platform was created through a user-centered design approach that involved end-users at every stage and is founded on the principles of cognitive behavioral therapy and the use of mind-body approaches. The core iCC features include daily symptom-tracking, personalized goal-setting to improve pain and functioning, and a toolbox of disease education and pain self-management strategies. A pilot study of iCC demonstrated feasibility of deploying the app among adolescents with chronic pain,[17] and has since been expanded to support pain self-management in youth and young adults with juvenile idiopathic arthritis,[18] sickle cell disease,[19] and post-operative pain, [20] with each adaptation reporting clinically meaningful reductions in pain intensity.

The use of mobile technology to address NF1 patients' needs is a promising strategy to expand access to pain self-management interventions. We therefore proposed to adapt the iCC platform. As a first step, we sought recommendations about the desired features and content that adults with NF1 and pain management experts considered important to include when adapting the iCC mobile health application as well as the perceived acceptability and barriers/facilitators to its use. We reasoned that the multiple perspectives of these individuals would yield a richer and more nuanced assessment of the desired features and content to include in the adaptation.

METHODS

Study Sample

The study involved two participant groups: Adults with NF1 (n = 32) and experts in the treatment and management of NF1-associated pain (n = 10). Inclusion criteria for the NF1 group were: (1) age 18-45 years with an NF1 diagnosis; (2) pain severity

aggregate score ≥3 on the Brief Pain Inventory-Short Form (BPI-SF)[21, 22] for pain experienced in the previous two weeks; (3) being proficient in English at the 5th grade level and competent to provide informed consent; (4) agreeing to audio-recording of the focus group session; and (5) residing in the U.S. The exclusion criteria consisted of: (1) moderate to severe cognitive deficits or (2) evidence of moderate to severe depression or anxiety, as assessed by the Patient Health Questionnaire (PHQ)[23] SCID-self-report (SCID-SR)[24] and Generalized Anxiety Disorder scale (GAD-7).[25] Inclusion criteria for the Expert group were: (1) providing healthcare to patients with NF1 (e.g., physicians, nurse practitioners, and psychologists) and (2) agreeing to audio-recording of the focus group session.

Patient and Public Involvement

Patients or the public were not involved in the design, or conduct, or reporting, or dissemination plans of this study.

Recruitment Procedures

Recruitment for the NF1 group consisted of posting an announcement briefly describing the study on the Facebook pages for two NF advocacy groups (NF Northeast and NF Network) and emailing all members of both group registries. Membership in both registries requires self-report of having received an NF diagnosis. Individuals interested in participating were directed to an online survey (www.qualtrics.com) that collected demographic and contact information as well as responses to the BPI, PHQ, GAD-7, and SCID-SR. The research team reviewed the results of these clinical

measures, and individuals meeting the inclusion requirements then received a list of focus group discussion (FGD) dates/times from which to select. Recruitment for the Expert group occurred via Website postings about the study on the US Association for the Study of Pain and the NF Network clinician page. Prior to beginning each FGD, participants were provided with a brief description of the study and informed of the voluntary nature of their participation. All participants provided verbal informed consent prior to participating in the FGD session.

After completing the FGD, participants in the NF1 and expert groups received a $25 and $100 gift card, respectively. The Institutional Review Board at Yale University approved all study materials and waived the need to obtain written consent.

Data Collection and Analysis

Each FGD session lasted approximately one hour and occurred via a secure, password-enforced video conference (www.zoom.com). Two members of the research team (FB, KL) with prior training in conducting focus groups jointly facilitated the sessions.

An interview guide was developed specifically for this study (see Supplementary Material). Only the NF1 group sessions began by asking whether participants owned smartphones. All participants were then asked their opinions about using a phone-based application to address NF1 pain management. A brief presentation then occurred that described the existing iCC mobile application and included four screen shots from the application. The remaining session time was guided by that description, the four screen shots, and the four domains included in the FGD guide: (1) perceived barriers and facilitators to using a mobile application to manage pain; (2) perceived acceptability and benefits of the proposed application; (3) contexts within which the application would most likely be used; and (4) recommendations for possible additions or modifications to the application. Demographic information for the NF1 group included age and gender, collected at eligibility screening, and for the Expert group included professional training/title, years in practice, and gender, collected at the beginning of each FGD. The audio-recorded transcripts were professionally transcribed verbatim.

An existing codebook from a prior iCC study[26] was modified for use in the current study. Members of the data analysis team (FB, KL, LG, JS, CL) met regularly to review transcripts and discuss codebook development. Interviews continued until data saturation had been achieved.[27, 28] The final codebook included 29 codes, three of which were new codes from the Expert group sessions. Acceptable inter-coder agreement was established after independently coding two transcripts. The remaining transcripts were then independently coded by both coders (FB, KL). The coding team continued to meet periodically to resolve coding discrepancies (through consensus), address coding questions, and further refine code definitions as needed. The coded transcripts were then entered into Dedoose (www.dedoose.com, version 8.3.47) for analysis. The coding and analysis process continued in an iterative fashion using thematic analysis.[29, 30] Negative instances where the data did not fit the existing themes were identified as part of the confirmability process.[31] Descriptive statistics were calculated for the demographic data. The entire research team reviewed all analytic findings.

RESULTS

A total of nine FGDs—six NF1 groups (n = 32) and three pain management expert groups (n = 10)—occurred between January and May 2020. Previous research has shown intrinsic motivation[32] to be the critical, first step in promoting future goal-oriented behaviors.[33] We therefore sought to identify themes that shaped motivation to use the proposed mobile application and generated a total of six themes across two levels (i.e., Individual and Application) as seen in the conceptual model in Figure 1. The themes of Lifestyle, Reasons for Using the Mobile App, and Concerns regarding Use of the Mobile Application were identified at the Individual Level. The App Level included the themes of Desired Content, Desired Features, and Format Considerations.

(Insert Figure here)

Description of the Study Sample

A total of 42 individuals participated in the study (Table 1). Overall, the NF1 group was generally young and predominantly female. Except for one participant, all owned smartphones. Most within the NF1 group were experiencing moderate pain at screening. The Expert group consisted entirely of individuals with post-graduate training in their respective fields and had, on average, over 19 years of professional experience. Similar to the NF1 group, the majority of expert participants were female.

Table 1. Characteristics of the Study Sample (N = 42)

	NF1 Group (n = 32)	Expert Group (n = 10)
Age (years)[1]	31.2 (4.9)	—
Range	18 - 39	
Gender[2]		
Male	6 (19%)	4 (40%)
Female	25 (78%)	6 (60%)
Other	1 (4%)	—
Aggregate Pain Severity[1,3]	4.8 (1.7)	—

Aggregate Pain Interference[1,4]	4.9 (2.6)	—
Professional Training/Title[2]		
Pain Psychologist	—	2 (20%)
Neurologist	—	5 (50%)
Primary Care Physician	—	2 (20%)
Neuro-oncologist	—	1 (10%)
Years in Practice[1]	—	19.2 (7.2)
Range		7 - 38

[1] Mean (S.D.)
[2] N (%)
[3] BPI-SF; 4 items for pain severity, possible range of scores is 0 - 10
[4] BPI-SF, 7 items for pain interference, possible range of scores is 0 – 10

Individual Level Themes

The Individual Level focused on the life situations of individuals with NF1 that were thought to influence motivation to use the proposed mobile application.

Table 2. Individual Level Themes and Quotes

Theme 1: Lifestyle	
Affordability/Accessibility sub-theme	Where I work there's certain applications that are blocked so it would be really a pain for lack of a better term for me to have to kind of remember everything and write it down later. (Participant 26, NF1 group)
	Unfortunately, in rural areas such as southwest Pennsylvania where I live or West Virginia a lot of people just don't have connections that are that strong sometimes. (Participant 24, NF1 group)
	There's always a lot of barriers in terms of access, economics, et cetera. (Participant 103, Expert group)
Time sub-theme	Sometimes I get up late and it's just something that I need to do every day but tend to forget if I'm hustling out the door or something. (Participant 7, NF1 group)
	I'm very bad about inputting stuff and keeping diaries and stuff because I have two toddlers at home, so that takes a lot of my time, too. (Participant 17, NF1 group)
	Other barriers are typically time… (Participant 106, Expert group)
Theme 2: Reasons for Using the Mobile Application	
Engagement sub-theme	I like apps that give me badges and awards for accomplishing my goals. (Participant 10, NF1 group)
	There has to be a way that they can, early in their treatment, start to see positive change, positive success

	so they get these little wins in the beginning and that's just the psychology of motivation and how you sort of really start to get that ball rolling. (Participant 104, Expert group)
Hope sub-theme	They have to be able to hold hope that they have the opportunity to be efficacious in improving the situation. (Participant 104, Expert group)
Theme 3: Concerns about Using the App	Who could potentially access that information? Could health insurance companies access…I'm worried about who has access to that and how they could potentially use it. (Participant 14, NF1 group)
	[Use of stars or points] can be very effective if they're tooled in the right way…but they [users] have to feel like that the goals are attainable. (Participant 104, female, Expert group)

Theme 1: Lifestyle (Individual Level)

Two sub-themes were identified within the Lifestyle theme (Table 2): Affordability/Accessibility and Time. The former focused on financial or other considerations that could influence ease of access. Both groups recognized that the mobile application needed to be "very cheap" or free and not consume much data (i.e., bottlenecking). Access issues related to geographic location or application use that depended upon external factors such as Internet signal strength or workplace access policies were also mentioned but were beyond what could be addressed within the application.

Time, the second sub-theme, focused on the busy lifestyles of most NF1 group members, usually involving parenting or work responsibilities. Both participant groups recognized that use of the application needed to easily fit into users' schedules. Barriers to its use included forgetting to access the application during busy times or when confronted with unexpected outside demands.

Theme 2: Reasons for Using the Mobile App (Individual Level)

This theme concerned potential factors thought to directly increase motivation to use the mobile application, primarily through rapid user engagement with program content and activities. It included two sub-themes: Engagement and Hope (Table 2).

The Engagement sub-theme focused on the fact that users would be more likely to use the application if they achieved improvements or successes early on. As one expert noted, “The ultimate aim is showing progress.” Both participant groups also believed that earning rewards (e.g., badges, points) can promote engagement with and continued use of the application. Participants often referred to this as “buy-in” and viewed it as critical to program uptake.

Similarly, instilling a sense of hope was seen as essential to promoting engagement and continued use of the program. Instilling hope was not considered to be limited to explicit messages of hope but could also result from individuals with NF1 recognizing improvements in function, quality of life, or having their NF1 pain validated by others.

Theme 3: Concerns regarding Use of the Mobile App (Individual Level)

The NF1 group seemed particularly concerned about privacy and access. They worried about who would have access to their health data and whether access to such data could jeopardize health insurance coverage or pose an unforeseen risk (Table 2). As previously mentioned in the Affordability/Accessibility sub-theme, participants were also concerned that Internet coverage or work restrictions could limit use of the application. Members of the Expert group were concerned that if goals were not realistic and achievable within an appropriate timeframe, some users might discontinue using the mobile application.

The Application Level

The three themes included at the Application Level focused on elements in the proposed mobile application that could increase users' motivation to use the product. The themes primarily concerned the content to be included and the functionality and features of the proposed product.

Theme 4: Desired Content (App Level)

Participants identified the important types of information about chronic pain and its management that is needed by individuals with NF1. It included the sub-themes of "Pain 101," Self-care, and Clinical Care (Table 3).

Table 3. Application Level Themes and Quotes for Desired Content and Desired Features

Theme 4: Desired Content	
"Pain 101" sub-theme	There's an abundance of research also on pain education as a pain management strategy…I do think the pain education piece and how pain works in the brain would be really great for NF patients. (Participant 101, Expert group)
	…how to work through and navigate the healthcare system, how to talk to healthcare providers about their pain and options. (Participant 106, Expert group)
	It's the specific exercises or to target or…to alleviate that specific pain. To have it be specific, broken down like a menu. Headaches? Do this. Sciatic pain? Do that. (Participant 2, NF1 group)
Self-care sub-theme	I think the mindfulness or meditation, yoga, breathing techniques would be extremely helpful. (Participant 26, NF1 group)
	…just getting them exposed to different complementary and alternative therapies has value, so even if you don't teach it in the app, if there was even a little module that was just like things you could try. (Participant 108, Expert group)
Clinical Care sub-theme	I did a study for NF with ACT, that's Acceptance and Commitment Treatment. I liked it because I think if you were going to do something with CBT [cognitive-behavior therapy], if you could incorporate some ACT as well, that would be great because to help you rethink about your pain might be, for me at least, it's helped me kind of restructure myself a little bit. (Participant 10, NF1 group)
	Cognitive behavioral therapy, evidence-based treatment for chronic pain involves a whole host of cognitive and behavioral strategies including pacing for pain and mindfulness, relaxation, catching

	catastrophic thoughts about pain and about the future, as well as lifestyle changes like nutrition and sleep hygiene. (Participant 101, Expert group)
Theme 5: Desired Features	
Tracking Capabilities sub-theme	I had to go through boxes of medical records to figure out and remember all the different doctors…so if there was a way to store some information… (Participant 3, NF1 group)
	I think one cool thing would be if there was an outline of a body that you could blow up and click on the part exactly that hurts and kind of color it in or shade it in or something" (Participant 19, NF1 group)
	I really struggle in striking the balance between having data on the pain but then also being able to do something about it…It's one thing if a patient came in and said, "Here's the graph, here's how much pain I've been having." I can look at that say, "Oh, that's rough. What are we going to do about that?" But if here's a graph then he or she also says here's the pain I was having and here's what else what was going on at that time and what I think may have contributed, then I may be able to offer some further intervention. (Participant 109, Expert group)
	I think tracking mood is hugely important. We know that anxiety and depression very commonly and understandably co-occur with chronic pain and medical conditions. We also know that mood and stress and anxiety and depression impact and amplify pain, so I definitely think it's important to assess mood and also to explain that mood and emotion is directly linked to the experience of pain. (Participant 101, Expert group)
Community sub-theme	…things where we could actually keep up with one another, talk to one another, just get things off our chest. I think that would be incredibly beneficial. (Participant 6, NF1 group)
	I think having positive or some opportunities to connect with other people that really understand the disease entity can be helpful too, as long as they're all working toward making gains and not just being really steeped and sad about the disease process. (Participant 104, Expert group)
	It was years before I ever met anybody else with NF and it's nice to know that there are other people out there…it would be nice on this app to be able to have maybe a comment section so that people can say, yup, I was there, or this is how I cope. (Participant 14, NF1 group)
	…people share incredible stuff on [message boards], so sharing pain may be nothing compared to sharing their personal stories about all their tumors and talking about a doctor. (Participant 107, Expert group)
External Resources sub-theme	I think of it as a way to maybe link certain maybe research articles or advice [that]… afterwards if you log a specific instance of pain and there's something that could pop up that you could read or if you log something that's maybe abnormal based on what you've logged in the past it could provide you some kind of feedback. (Participant 6, NF1 group)
	We use [name of app] …And it's been really helpful for things like CRPS [complex regional pain syndrome], potentially amputation pain, and so it's part of cognitive retraining for the disorganized cognitive input from a missing limb or a dysfunctional limb. (Participant 105, Expert group)

Future Features sub-theme	One thing I'd like to see for the physical activity, if it could import data from say [names of activity apps] or whatever if you use one of those apps, it would be one less thing for the user to do. (Participant 20, NF1 group)
	I'm a software developer myself so I want my app to be able to function at least as good as it can with no Internet connection and automatically update the database when I get connection. (Participant 24, NF1 group)
	One of the apps I used to use was for water and as you drank water it grew a plant. Well, maybe something interactive based on the age ranges of who might use it. (Participant 9, NF1 group)
	…if [patients] can print out a graph or somehow send the graphical representation that we've like scanned that into the medical record, that's been helpful (Participant 108, Expert group)
	I think one really important feature that a lot of other applications that I use lack is integration with MyChart and other things that my doctors would have access to so that everybody can just see it rather than having to go back and forth with all the different doctors (Participant 26, NF1 group)

The experts believed that accurate knowledge about the topic of pain was important in motivating individuals to use the mobile application— "a very essential piece of this work, not sufficient, but necessary" and referred to it as "Pain 101" information. This included information on the physiological basis of pain, neuropathic and nociplastic pain, the difference between acute and chronic pain, and NF1-specific pain. They also noted that users should receive instruction about how to successfully navigate the healthcare system and effectively interact with their healthcare providers.

By contrast, the NF1 group appeared most interested in learning about symptom recognition, recommended analgesics, and strategies for targeting specific types of pain.

The Self-care sub-theme dealt with pain treatment approaches potential users could learn and then do independently or in group settings. Both participant groups offered many suggestions about possible pain mitigation strategies. These could be divided into two types of self-care strategies: physical and psychobehavioral. The former, endorsed by both groups, included movement-based activities such as martial arts, yoga or "grounding exercises," and pacing. Only the Expert group suggested improvement of sleep hygiene and use of heat and ice as physical approaches to self-management of NF1 pain. Both groups suggested psychobehavioral approaches (i.e., those requiring only brief training with a qualified professional) such as deep, circular, or meditational breathing and music or aroma therapy.

The final sub-theme, Clinical Care, focused on pain treatments that required meeting with a trained professional for longer periods than typically required in Self-care approaches. Yet, similar to the Self-care sub-theme, the treatments could be divided into physical and psychobehavioral approaches. Only the expert group identified physical approaches such as physical therapy or nutrition care. Longer term psychobehavioral approaches included cognitive behavior therapy, acceptance and commitment therapy, guided imagery, biofeedback, and mindfulness training.

Theme 5: Desired Features (App Level)

The Desired Features theme included four sub-themes: (1) Tracking capabilities, (2) Community, (3) External Resources, and (4) Future Features. This theme focused on the functionality of the mobile application and the desire for links to external resources (Table 3).

Tracking of various events and information was considered by both participant groups to be an important feature of the mobile application. It included tracking and monitoring pain level, mood, activity, sleep, and stress, all considered to be important potential pain triggers. Both groups also believed that maintaining notes or a diary was important but for somewhat different reasons. The NF1 group viewed this feature as a potential memory aid that could enable patients to “talk about [pain symptoms] more intelligently to a provider.” It could serve as a reminder for exercising, logging information in the application, or to track meditation and physical activity. The NF1 group also expressed interest in recording their surgical and medication histories and having the capability of tracking pain details such as onset, location, quality, and level of pain.

By contrast, the Expert group appeared to be interested in a less detailed approach to tracking pain, albeit sufficiently detailed to be able to identify associations between pain and such things as mood, activity, or treatment efficacy.

Both participant groups were enthusiastic about creating an online community that could serve as a resource for socioemotional support and NF1-relevant information (e.g., local events, providers). Members of the Expert group cautioned that it should not be a venue for complaining about having the disease, however. The NF1 group offered many suggestions about the format (e.g., group size, age restrictions, creation of a buddy system), although none emerged as the preferred format. Both groups also agreed that sharing experiences could help normalize having NF1 and help individuals feel less isolated.

Participants believed that external resources such as links to other mobile applications could be helpful to NF1 patients. The NF1 group was interested in links to relevant articles and treatment guidelines. The Expert group suggested links that could assist individuals with NF1 in self-management of their disease, including relaxation and pain sites.

Both groups identified some features that were currently not feasible to include in the mobile application, as they would require extensive resources, negotiations with other applications, or lengthy modifications to the current iCC. They can be considered for future modifications to the application. Suggestions included automatic data upload at the next sign-in to make it more convenient for users whose Internet access is blocked at work or in certain geographical locations, printable reports, 24/7 access to medical advice, and a visual graphic representation of progress toward a specific goal.

Other suggestions, primarily from the NF1 group, were currently unrealistic to implement because they require formalizing agreements with healthcare institutions. For example, they mentioned wanting the capability to automatically share information with healthcare providers or access existing x-rays when at another location.

Theme 6: Format Considerations (App Level)

The final theme included two sub-themes (Table 4): User Interface and Presentation/Tone. The first addressed issues that would increase users' ease of use, and the second focused on the visual appeal and tone of the content presented.

Table 4. Application Level Theme and Quotes for Format Considerations

Theme 6: Format Considerations	
User Interface sub-theme	I need consistency. Kind of like if you're going to do updates and stuff like that, it needs to not be completely reconfigured. (Participant 1, NF1 group)
	If I can add in my own entries along with that, that would motivate me to use it more. (Participant 18, NF1 group)
	…then you want a very simple front page and then links or tabs. (Participant 105, Expert group)
Presentation/Tone sub-theme	I think having some visuals as well would be useful. Yeah, visual, not horrid looking. (Participant 1, NF1 group)
	I would want it to be simple and fun and use language that I use, not language a doctor would use. I can look up words, but sometimes I don't want to. (Participant 4, NF1 group)
	I think considering color is important…We didn't really talk about that but, the whole visual appeal. Because if it's not appealing or if

	it's going to make a person feel worse then that's counter, then that's going against what [the goal of the application is]. (Participant 102, Expert group)
	When I was looking at your app, it made me think about music and how that can be useful for people's state of mind, which you can't separate the body from the mind so I think that would be great in chronic pain. (Participant 103, Expert group)

Both groups understood the importance of User Interface, that motivation to use the mobile application could be heavily influenced by its ease of use. The Expert group suggested having explicit instructions on how to navigate and use the application. Participants from both groups recommended tailoring the application to users' specific preferences on pain-tracking measures and format (e.g., numeric vs. figural, pain descriptors) and including an "Other" option so users could further qualify their responses to forced-choice questions. They also identified features to enhance time-efficiency such as auto-population of key information, use of drop-down menus, and consistency in format and content when updating.

Critical elements in motivating individuals to use the mobile application were Tone and Presentation, the final sub-theme. Participants described the ideal application as being "approachable and digestible" and "simple and fun." Visual appeal was important, noting that the font and use of color could influence use. Careful selection of NF1 photographs and language could also be used to set an appropriate tone. Members of the NF1 group believed that daily tips or inspiring quotes could enhance engagement, and experts recommended varying the messages to keep the content fresh. Both groups agreed that music could help set a positive tone as well.

DISCUSSION

Few studies have examined the perceived benefits, desired content and features, or efficacy of mobile health applications on coping with NF1-related chronic pain. To our knowledge, the current study is the only one to examine some of these issues from the perspectives of the targeted end-users and pain management experts. We believe that this is essential information to ensure that the product is scientifically accurate and relevant while also satisfying targeted end-users' needs and preferences.

Both groups were enthusiastic about the prospect of adapting the iCC mobile application for individuals with NF1 with chronic pain. They believed that tailoring the program to accommodate users' lifestyle needs and affordability, incentivizing its use, fostering an NF1 community support group, and using appropriate and acceptable tone and format features will encourage uptake and continued use of the mobile application. In addition, offering NF1 and coping skills information and an online support community in a single mobile application renders it both convenient and comprehensive in scope.

Participants believed that flexibility in fitting into potential users' current situation was important. They noted that the application should be affordable, easy to use, and convenient; it should fit users' schedules, efficiently capture the desired data, and be modifiable as users' life situations change. These findings support previous research on features needed to ensure uptake and ease of use of the mobile application[34, 35] as well as health-related mobile application users' privacy and security concerns.[36, 37]

With respect to the issue of promoting sustained use of the application, participants from both groups noted the importance of including realistic and achievable goals. Early improvements might involve pain reduction or changes in pain attitudes that could improve mood, function, or quality of life. Consistent with the notion of contingency management,[38, 39] participants identified strategies involving points, levels, prizes, or well-established reinforcement schedules to incentivize continued use. These findings suggest the importance of considering contingency management when developing an application to promote health behaviors.[40,41]

Regarding the pain symptom tracking feature, the two groups had somewhat different views on what type of pain data are most desired. The NF1 group appeared interested in gathering as detailed and nuanced pain information as possible. By contrast, the Expert group appeared more interested in tracking information that might identify potential associations between pain, mood, nutrition, activity, and stress levels; they believed that noting these patterns would be more useful than tracking pain intensity, duration, quality, and directionality. Consistent with previous research,[42] the two participant groups expressed different perspectives on and desires for tracking chronic pain. The data suggest that to accommodate these disparate views, the pain tracking feature should permit simultaneous tracking of several constructs (e.g., pain, mood, activity) while also providing a flexible format so that patients can select the level of detail and pain metrics they prefer. One cautionary note, however, would be to avoid the potential for users to become fixated on their pain symptoms to the detriment of recognizing the associations between their pain and the other constructs. Based on input from our Expert group, we plan to develop an app that is engaging without over-focusing on tracking or complaining about symptoms and subsequently assess its effectiveness and potential positive/negative effects.

Community support, particularly for individuals with rare diseases such as NF1, can be an important coping resource.[43,44] Previous research on online community support platforms for individuals with rare diseases has noted that they are used for both informational and socioemotional support purposes.[45, 46] The value of community support has been identified within the context of cultural and racial socialization,[47, 48] and the study corroborates these findings. Both groups noted how the creation of an online NF1 community would provide users the opportunity to share and thereby validate their experiences with the disease and chronic pain, provide socioemotional encouragement, and potentially motivate continued use of the mobile application.

The study supported previous findings about visual appeal, tone, ease of use, instructions on effectively navigating app options, tailoring mHealth interventions to personal needs and preferences, and the use of appropriate visuals.[49, 50] This last feature is particularly important with a potentially disfiguring disease such as NF1. A novel finding from this study, however, is the importance of establishing an upbeat tone that instills hope.

We note several study limitations. First, the findings may have limited transferability, given the modest racial and ethnic diversity of the study sample. Second, lack of external confirmation of self-reported NF1 diagnoses may have limited the credibility of the results. Third, the myriad suggestions proffered may be attributed to social desirability wherein participants sought to identify as many options as possible in order to “please” the group facilitators. Nonetheless, these options made possible a deeper analysis of potential format and content considerations when adapting the iCC application. Finally, virtual FGDs limit the ability to observe subtle, in-person dynamics during discussions. We chose this format, however, prior to the onset of the COVID-19 pandemic, based upon our desire to collect data in a time- and cost-efficient manner across diverse geographic regions and living situations.

Conclusions

The study findings suggest that adapting the iCC mobile application for use in the self-management of NF1-associated chronic pain was seen by adults with NF1 and chronic pain experts as a welcomed addition to the armamentarium of NF1 treatment resources. Careful attention should be paid to the creation of a community support group option as well as program content, format, and ability to tailor it to users' specific needs.

Acknowledgements: We are indebted to Karen Peluso, Director of NF Northeast and Kim Bischoff, Director of NF Network for their help and commitment. We also thank the study participants for sharing with us their time and insights.

Funding: This work was supported by the U.S. Army Medical Research Materiel Command endorsed by the U.S. Army, through the Congressionally Directed Medical Research Programs' Neurofibromatosis Research Program under Award No. W81XWH-19-1-0618.

Competing Interests Statement: None declared.

Author Contributions:

LG- Manuscript preparation and review, study design and data analysis, development of interview guide

KL- Daily oversight of project, focus group facilitation, data management and analysis, manuscript preparation and review

CL- Development of interview guide, manuscript preparation and review

JS- Development of interview guide, manuscript preparation and review

WZ- Supervision of the project, manuscript review

SB- Supervision of the project, manuscript review

FB- Study design and data analysis, led the focus groups, development of interview guide, manuscript preparation and review, oversight for the general conduct of this study

Data Sharing Statement: Requests to receive the qualitative data should be addressed to the Corresponding Author. Upon review of the request, the data may be made available.

Ethics Statement: The Human Subjects Committee of the Yale Human Research Protection Program of Institutional Review Boards at Yale University approved Protocol #2000025437.

Figure Legend: Six themes identified within the data were thought to shape motivation to use the mobile application. Three could be attributed to personal characteristics of the targeted end-users and were thought to directly influence motivation. The remaining three focused on content and qualities of the application itself and were thought to indirectly influence motivation to use the application.

REFERENCES

1. Anderson JL, Gutmann DH. Neurofibromatosis type 1. *Handb Clin Neurol* 2015;132:75-86. PMID: 26564071. doi: 10.1016/B978-0-444-62702-5.00004-4.
2. Kodra Y, Giustini S, Divona L, Porciello R, Calvieri S, Wolkenstein P, et al. Health-related quality of life in patients with neurofibromatosis type 1. A survey of 129 Italian patients. *Dermatology* 2009;218:215-20. PMID: 19088462. doi: 10.1159/000187594.
3. Radtke HB, Sebold CD, Allison C, Haidle JL, Schneider G. Neurofibromatosis type 1 in genetic counseling practice: recommendations of the National Society of Genetic Counselors. *J Genet Couns* 2007;16:387-407. PMID: 17636453. doi: 10.1007/s10897-007-9101-8.
4. Tonsgard JH. Clinical manifestations and management of neurofibromatosis type 1. *Semin Pediatr Neurol* 2006;13:2-7. PMID: 16818170. doi: 10.1016/j.spen.2006.01.005.
5. Brems H, Beert E, de Ravel T, Legius E. Mechanisms in the pathogenesis of malignant tumours in neurofibromatosis type 1. *Lancet Oncol* 2009;10:508-15. PMID: 19410195. doi: 10.1016/s1470-2045(09)70033-6.
6. North KN, Riccardi V, Samango-Sprouse C, Ferner R, Moore B, Legius E, et al. Cognitive function and academic performance in neurofibromatosis. 1: consensus statement from the NF1 Cognitive Disorders Task Force. *Neurology* 1997;48:1121-7. PMID: 9109916. doi: 10.1212/wnl.48.4.1121.
7. Vranceanu A-M, Merker VL, Park E, Plotkin SR. Quality of life among adult patients with neurofibromatosis 1, neurofibromatosis 2 and schwannomatosis: a systematic review of the literature. *J Neurooncol* 2013;114:257-62. doi: 10.1007/s11060-013-1195-2.
8. Sabatini C, Milani D, Menni F, Tadini G, Esposito S. Treatment of neurofibromatosis type 1. *Curr Treat Options Neurol* 2015;17(6):355. PMID: 25917340. doi: 10.1007/s11940-015-0355-4.
9. Buono FD, Grau LE, Sprong ME, Morford KL, Johnson KJ, Gutmann DH. Pain symptomology, functional impact, and treatment of people with Neurofibromatosis type 1. J *Pain Res* 2019;12:2555-61. PMID: 31692483. doi: 10.2147/jpr.S209540.
10. Martin S, Wolters PL, Toledo-Tamula MA, Schmitt SN, Baldwin A, Starosta A, et al. Acceptance and commitment therapy in youth with neurofibromatosis type 1 (NF1) and chronic pain and their parents: A pilot study of feasibility and preliminary efficacy. *Am J Med Genet A* 2016;170:1462-70. PMID: 27021207. doi: 10.1002/ajmg.a.37623.
11. Ferrara G, Kim J, Lin S, Hua J, Seto E. A Focused Review of Smartphone Diet-Tracking Apps: Usability, Functionality, Coherence With Behavior Change Theory, and Comparative Validity of Nutrient Intake and Energy Estimates. *JMIR Mhealth Uhealth* 2019;7:e9232. PMID: 31102369. doi: 10.2196/mhealth.9232.
12. Birkhoff SD, Smeltzer SC. Perceptions of Smartphone User-Centered Mobile Health Tracking Apps Across Various Chronic Illness Populations: An Integrative

Review. *J Nurs Scholarsh* 2017;49:371-8. PMID: 28605151. doi: 10.1111/jnu.12298.

13. Sarkar U, Gourley GI, Lyles CR, Tieu L, Clarity C, Newmark L, et al. Usability of Commercially Available Mobile Applications for Diverse Patients. *J Gen Intern Med* 2016;31:1417-26. PMID: 27418347. doi: 10.1007/s11606-016-3771-6.
14. Whitehead L, Seaton P. The Effectiveness of Self-Management Mobile Phone and Tablet Apps in Long-term Condition Management: A Systematic Review. *J Med Internet Res* 2016;18:e97. PMID: 27185295. doi: 10.2196/jmir.4883.
15. Stinson JN, Lalloo C, Harris L, Isaac L, Campbell F, Brown S, et al. iCanCope with Pain™: User-centred design of a web- and mobile-based self-management program for youth with chronic pain based on identified health care needs. *Pain Res Manag* 2014;19:257-65. PMID: 25000507. doi: 10.1155/2014/935278.
16. Stinson JN, White M, Isaac L, Campbell F, Brown S, Ruskin D, et al. Understanding the information and service needs of young adults with chronic pain: perspectives of young adults and their providers. *Clin J Pain* 2013;29:600-12. PMID: 23328333. doi: 10.1097/AJP.0b013e31826dce65.
17. Lalloo C, Hundert A, Harris L, Pham Q, Campbell F, Chorney J, et al. Capturing Daily Disease Experiences of Adolescents With Chronic Pain: mHealth-Mediated Symptom Tracking. *JMIR Mhealth Uhealth* 2019;7:e11838. PMID: 30664472. doi: 10.2196/11838.
18. Lalloo C, Harris LR, Hundert AS, Berard R, Cafazzo J, Connelly M, et al. The iCanCope pain self-management application for adolescents with juvenile idiopathic arthritis: a pilot randomized controlled trial. *Rheumatology (Oxford)* 2021;60:196-206. PMID: 32613229. doi: 10.1093/rheumatology/keaa178.
19. Palermo TM, Zempsky WT, Dampier CD, Lalloo C, Hundert AS, Murphy LK, et al. iCanCope with Sickle Cell Pain: Design of a randomized controlled trial of a smartphone and web-based pain self-management program for youth with sickle cell disease. *Contemp Clin Trials* 2018;74:88-96. PMID: 30316999. doi: 10.1016/j.cct.2018.10.006.
20. Birnie KA, Campbell F, Nguyen C, Lalloo C, Tsimicalis A, Matava C, et al. iCanCope PostOp: User-Centered Design of a Smartphone-Based App for Self-Management of Postoperative Pain in Children and Adolescents. *JMIR Form Res* 2019;3:e12028. PMID: 31008704. doi: 10.2196/12028.
21. Cleeland CS, Ryan KM. Pain assessment: global use of the Brief Pain Inventory. *Ann Acad Med Singap* 1994;23:129-38. PMID: 8080219.
22. Palos GR, Mendoza TR, Mobley GM, Cantor SB, Cleeland CS. Asking the community about cutpoints used to describe mild, moderate, and severe pain. *J Pain* 2006;7:49-56.
23. Kroenke K, Spitzer RL, Williams JB. The PHQ-9: validity of a brief depression severity measure. *J Gen Intern Med* 2001;16:606-13. PMID: 11556941. doi: 10.1046/j.1525-1497.2001.016009606.x.

24. Brodey B, Purcell SE, Rhea K, Maier P, First M, Zweede L, et al. Rapid and Accurate Behavioral Health Diagnostic Screening: Initial Validation Study of a Web-Based, Self-Report Tool (the SAGE-SR). *J Med Internet Res* 2018;20:e108. PMID: 29572204. doi: 10.2196/jmir.9428.

25. Spitzer RL, Kroenke K, Williams JB, Lowe B. A brief measure for assessing generalized anxiety disorder: the GAD-7. *Arch Intern Med* 2006;166):1092-7. PMID: 16717171. doi: 10.1001/archinte.166.10.1092.

26. Ahola Kohut S, LeBlanc C, O'Leary K, McPherson AC, McCarthy E, Nguyen C, et al. The internet as a source of support for youth with chronic conditions: A qualitative study. *Child Care Health Dev* 2018;44:212-20. PMID: 29082537. doi: 10.1111/cch.12535.

27. Guest G, Bunce A, Johnson L. How Many Interviews Are Enough?:An Experiment with Data Saturation and Variability. *Field Methods* 2006;18:59-82. doi: 10.1177/1525822x05279903.

28. Hennink MM, Kaiser BN, Marconi VC. Code Saturation Versus Meaning Saturation: How Many Interviews Are Enough? *Qual Health Res* 2017;27:591-608. PMID: 27670770. doi: 10.1177/1049732316665344.

29. Braun V, Clarke V. Using thematic analysis in psychology. Qualitative Research in Psychology. 2006 2006/01/01;3(2):77-101. doi: 10.1191/1478088706qp063oa.

30. Braun V, Clarke V. Successful qualitative research: A practical guide for beginners: sage; 2013. ISBN: 1446289516.

31. Henwood KL, Pidgeon NF. Qualitative research and psychological theorizing. Br J Psychol. 1992 Feb;83 (Pt 1):97-111. PMID: 1559146. doi: 10.1111/j.2044-8295.1992.tb02426.x.

32. Deci EL, Ryan RM. Intrinsic Motivation and Self-Determination in Human Behavior.1985. New York:Springer Science+Business Media. ISBN: 978-1-4899-2273-1.

33. Ajzen I. From Intentions to Actions: A Theory of Planned Behavior. In: Kuhl J, Beckmann J, editors. Action Control: From Cognition to Behavior. Berlin:Springer-Verlag Berlin Heidelberg; 1985. p. 11-39. ISBN: 978-3-642-69746-3

34. O'Brien T, Rosenthal A. Preferred Features in Mobile Health Applications for Kidney Transplant Recipients: A Qualitative Approach. *Nephrol Nurs J* 2020;47:529-36. [published Online First: 2020/12/31]

35. Zapata BC, Fernández-Alemán JL, Idri A, et al. Empirical Studies on Usability of mHealth Apps: A Systematic Literature Review. *J Med Syst* 2015;39:1. doi: 10.1007/s10916-014-0182-2

36. Martínez-Pérez B, de la Torre-Díez I, López-Coronado M. Privacy and security in mobile health apps: a review and recommendations. *J Med Syst* 2015;39:181. doi: 10.1007/s10916-014-0181-3

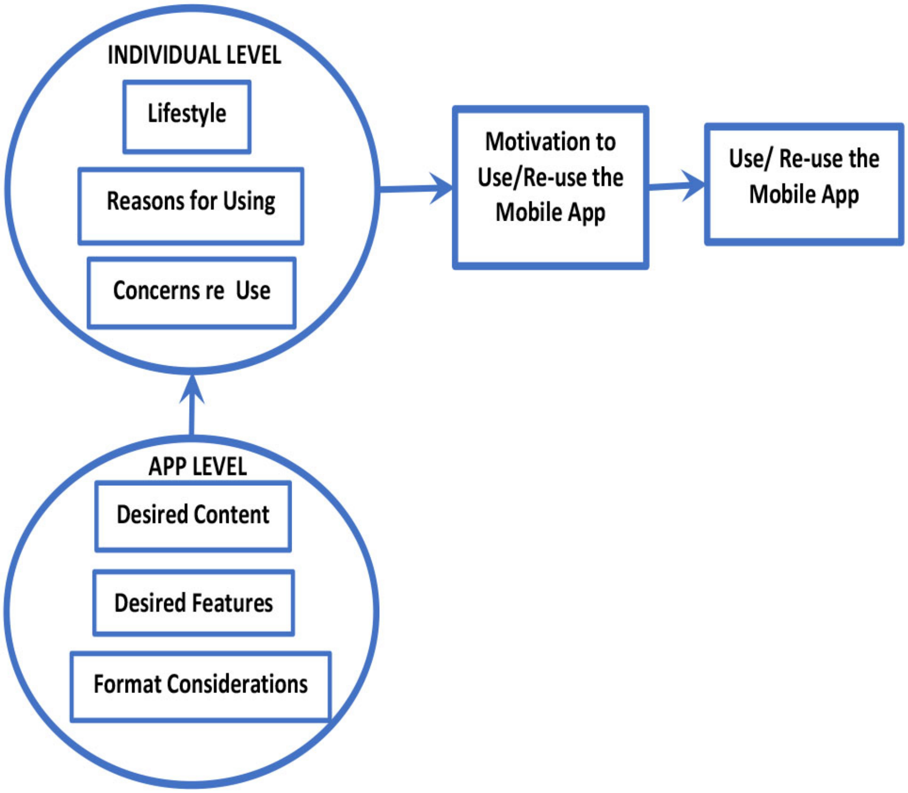
37. Agarwal P, Gordon D, Griffith J, Kithulegoda N, Witteman HO, Sacha Bhatia R, et al. Assessing the quality of mobile applications in chronic disease management: a

scoping review. *NPJ Digit Med* 2021;4:46. PMID: 33692488. doi: 10.1038/s41746-021-00410-x.

38. Higgins ST, Petry NM. Contingency management. Incentives for sobriety. *Alcohol Res Health* 1999;23:122-7. PMID: 10890806.
39. Higgins ST, Silverman K. Motivating behavior change among illicit-drug abusers: Research on contingency management interventions. Washington, DC, US: American Psychological Association; 1999. ISBN: 1-55798-570-7.
40. Máca V, Ščasný M, Zvěřinová I, et al. Incentivizing Commuter Cycling by Financial and Non-Financial Rewards. *Int J Environ Res Public Health* 2020;17:6033 doi: 10.3390/ijerph17176033
41. Carpenter VL, Hertzberg JS, Kirby AC, et al. Multicomponent smoking cessation treatment including mobile contingency management in homeless veterans. *J Clin Psychiatry* 2015;76:959-64. doi: 10.4088/JCP.14m09053
42. Kovačević I, Kogler VM, Turković TM, Dunkić LF, Ivanec Ž, Petek D. Self-care of chronic musculoskeletal pain - experiences and attitudes of patients and health care providers. *BMC Musculoskelet Disord* 2018;19:76. PMID: 29514616. doi: 10.1186/s12891-018-1997-7.
43. Martin S, Struemph KL, Poblete A, et al. An Internet support group for parents of children with neurofibromatosis type 1: a qualitative analysis. *J Community Genet* 2018;9:327-34. doi: 10.1007/s12687-018-0360-x
44. Martin S, Wolters PL, Baldwin A, et al. Attitudes about internet support groups among adolescents and young adults with neurofibromatosis type 1 and their parents. *J Genet Couns* 2014;23:796-804. doi: 10.1007/s10897-014-9688-5
45. Lasker JN, Sogolow ED, Sharim RR. The role of an online community for people with a rare disease: content analysis of messages posted on a primary biliary cirrhosis mailinglist. *J Med Internet Res* 2005;7:e10. PMID: 15829472. doi: 10.2196/jmir.7.1.e10.
46. Doyle M. Peer Support and Mentorship in a US Rare Disease Community: Findings from the Cystinosis in Emerging Adulthood Study. *Patient* 2015;8:65-73. PMID: 25231828. doi: 10.1007/s40271-014-0085-9.
47. Litchman ML, Edelman LS, Donaldson GW. Effect of Diabetes Online Community Engagement on Health Indicators: Cross-Sectional Study. *JMIR Diabetes* 2018;3:e8. doi: 10.2196/diabetes.8603
48. Young SD, Holloway I, Jaganath D, et al. Project HOPE: online social network changes in an HIV prevention randomized controlled trial for African American and Latino men who have sex with men. *Am J Public Health* 2014;104:1707-12. doi: 10.2105/ajph.2014.301992
49. Llorens-Vernet P, Miró J. Standards for Mobile Health-Related Apps: Systematic Review and Development of a Guide. *JMIR Mhealth Uhealth* 2020;8:e13057. PMID: 32130169. doi: 10.2196/13057.
50. Woods L, Duff J, Cummings E, Walker K. Evaluating the Development Processes of Consumer mHealth Interventions for Chronic Condition Self-management: A

Scoping Review. *Comput Inform Nurs* 2019;37:373-85. PMID: 31135468. doi: 10.1097/cin.0000000000000528.

Figure 1 Conceptual Framework and Themes for Motivation to Use the Mobile Application


INDIVIDUAL LEVEL
Lifestyle
Reasons for Using
Concerns re Use
Motivation to Use/Re-use the Mobile App
Use/ Re-use the Mobile App
APP LEVEL
Desired Content
Desired Features
Format Considerations

Legend: Six themes identified within the data were thought to shape motivation to use the mobile application. Three could be attributed to personal characteristics of the targeted end-users and were thought to directly influence motivation. The remaining three focused on content and qualities of the application itself and were thought to indirectly influence motivation to use the application.

90x90mm (300 x 300 DPI)

SRQR Reporting checklist for qualitative study.

Instructions to authors

Complete this checklist by entering the page numbers from your manuscript where readers will find each of the items listed below. SRQR reporting guidelines: O'Brien BC, Harris IB, Beckman TJ, Reed DA, Cook DA. Standards for reporting qualitative research: a synthesis of recommendations. Acad Med. 2014;89(9):1245-1251.

		Reporting Item	Page Number
Title			
	#1	Concise description of the nature and topic of the study identifying the study as qualitative or indicating the approach (e.g. ethnography, grounded theory) or data collection methods (e.g. interview, focus group) is recommended	Title
Abstract			
	#2	Summary of the key elements of the study using the abstract format of the intended publication; typically includes background, purpose, methods, results and conclusions	Abstract
Introduction			
Problem formulation	#3	Description and signifcance of the problem / phenomenon studied: review of relevant theory and empirical work; problem statement	Introduction, Paragraphs 1-4
Purpose or research question	#4	Purpose of the study and specific objectives or questions	Introduction Para 5
Methods			
Qualitative approach and research paradigm	#5	Qualitative approach (e.g. ethnography, grounded theory, case study, phenomenolgy, narrative research) and guiding theory if appropriate; identifying the research paradigm (e.g. postpositivist, constructivist / interpretivist) is also recommended; rationale. The rationale should briefly discuss the justification for choosing that theory, approach, method or technique rather than other options available; the assumptions and limitations implicit in those choices and how those choices influence study conclusions and transferability. As appropriate the rationale for several items might be discussed together.	Introduction – Para 5 Methods – Para 1 - 7 Methods: Para 5, 7
Researcher characteristics and reflexivity	#6	Researchers' characteristics that may influence the research, including personal attributes, qualifications / experience, relationship with participants, assumptions and / or presuppositions; potential or actual interaction between researchers' characteristics and the research questions, approach, methods, results and / or transferability	Methods: Recruitment Procedures
Context	#7	Setting / site and salient contextual factors; rationale	
Sampling strategy	#8	How and why research participants, documents, or events were selected; criteria for deciding when no further sampling was necessary (e.g. sampling saturation); rationale	
Ethical issues pertaining to human subjects	#9	Documentation of approval by an appropriate ethics review board and participant consent, or explanation for lack thereof; other confidentiality and data security issues	Data Collection and Analysis Para 3
Data collection methods	#10	Types of data collected; details of data collection procedures including (as appropriate) start and stop dates of data collection and analysis, iterative process, triangulation of sources / methods, and modification of procedures in response to evolving study findings; rationale	Recruitment Procedure Para 2
Data collection	#11	Description of instruments (e.g. interview guides,	Data Collection Para 1-3

instruments and technologies		questionnaires) and devices (e.g. audio recorders) used for data collection; if / how the instruments(s) changed over the course of the study	Data Collection Para 2
Units of study	#12	Number and relevant characteristics of participants, documents, or events included in the study; level of participation (could be reported in results)	Description of Study Sample and Table 1
Data processing	#13	Methods for processing data prior to and during analysis, including transcription, data entry, data management and security, verification of data integrity, data coding, and anonymisation / deidentification of excerpts	Data Collection and Analysis Para 1 - 3
Data analysis	#14	Process by which inferences, themes, etc. were identified and developed, including the researchers involved in data analysis; usually references a specific paradigm or approach; rationale	Data Collection and Analysis Para 3
Techniques to enhance trustworthiness	#15	Techniques to enhance trustworthiness and credibility of data analysis (e.g. member checking, audit trail, triangulation); rationale	Patient and Public Involvement; Data Collection and
Results/findings			
Syntheses and interpretation	#16	Main findings (e.g. interpretations, inferences, and themes); might include development of a theory or model, or integration with prior research or theory	Results Para 1 + results
Links to empirical data	#17	Evidence (e.g. quotes, field notes, text excerpts, photographs) to substantiate analytic findings	Results
Discussion			
Intergration with prior work, implications, transferability and contribution(s) to the field	#18	Short summary of main findings; explanation of how findings and conclusions connect to, support, elaborate on, or challenge conclusions of earlier scholarship; discussion of scope of application / generalizability; identification of unique contributions(s) to scholarship in a discipline or field	Conclusions
Limitations	#19	Trustworthiness and limitations of findings	Discussion: Para 8
Other			
Conflicts of interest	#20	Potential sources of influence of perceived influence on study conduct and conclusions; how these were managed	Back materials
Funding	#21	Sources of funding and other support; role of funders in data collection, interpretation and reporting	Funding; Author Contributions

None The SRQR checklist is distributed with permission of Wolters Kluwer © 2014 by the Association of American Medical Colleges. This checklist can be completed online using https://www.goodreports.org/, a tool made by the EQUATOR Network in collaboration with Penelope.ai

BMJ Open

Perspectives on Adapting a Mobile Application for Pain Self-Management in Neurofibromatosis Type 1: Results of Online Focus Group Discussions with Individuals Living with Neurofibromatosis Type 1 and Pain Management Experts

Journal:	*BMJ Open*
Manuscript ID	bmjopen-2021-056692.R2
Article Type:	Original research
Date Submitted by the Author:	20-May-2022
Complete List of Authors:	Grau, Lauretta; Yale University Yale School of Public Health, Department of Epidemiology of Microbial Diseases Larkin, Kaitlyn; Yale School of Medicine, Department of Psychiatry Lalloo, Chitra; The Hospital for Sick Children, Child Health Evaluative Sciences; University of Toronto, Institute of Health Policy, Management & Evaluation Stinson, Jennifer; Toronto SickKids, Child Health Evaluative Sciences; University of Toronto, Institute of Health Policy, Management & Evaluation Zempsky, William; University of Connecticut School of Medicine, Department of Pediatrics and Nursing; Connecticut Children's Medical Center, Division of Pain and Palliative Care Ball, Samuel; Yale School of Medicine, Department of Psychiatry Buono, Frank; Yale School of Medicine, Department of Psychiatry
Primary Subject Heading:	Complementary medicine
Secondary Subject Heading:	Neurology
Keywords:	COMPLEMENTARY MEDICINE, Adult neurology < NEUROLOGY, Neurogenetics < NEUROLOGY, PAIN MANAGEMENT, QUALITATIVE RESEARCH, Neurological pain < NEUROLOGY

SCHOLARONE™
Manuscripts

I, the Submitting Author has the right to grant and does grant on behalf of all authors of the Work (as defined in the below author licence), an exclusive licence and/or a non-exclusive licence for contributions from authors who are: i) UK Crown employees; ii) where BMJ has agreed a CC-BY licence shall apply, and/or iii) in accordance with the terms applicable for US Federal Government officers or employees acting as part of their official duties; on a worldwide, perpetual, irrevocable, royalty-free basis to BMJ Publishing Group Ltd ("BMJ") its licensees and where the relevant Journal is co-owned by BMJ to the co-owners of the Journal, to publish the Work in this journal and any other BMJ products and to exploit all rights, as set out in our licence.

The Submitting Author accepts and understands that any supply made under these terms is made by BMJ to the Submitting Author unless you are acting as an employee on behalf of your employer or a postgraduate student of an affiliated institution which is paying any applicable article publishing charge ("APC") for Open Access articles. Where the Submitting Author wishes to make the Work available on an Open Access basis (and intends to pay the relevant APC), the terms of reuse of such Open Access shall be governed by a Creative Commons licence – details of these licences and which Creative Commons licence will apply to this Work are set out in our licence referred to above.

Other than as permitted in any relevant BMJ Author's Self Archiving Policies, I confirm this Work has not been accepted for publication elsewhere, is not being considered for publication elsewhere and does not duplicate material already published. I confirm all authors consent to publication of this Work and authorise the granting of this licence.

Perspectives on Adapting a Mobile Application for Pain Self-Management in Neurofibromatosis Type 1: Results of Online Focus Group Discussions with Individuals Living with Neurofibromatosis Type 1 and Pain Management Experts

Lauretta E. Grau, PhD[1], Kaitlyn Larkin, BA[2], Chitra Lalloo, PhD[3,4], Jennifer N. Stinson, RN, PhD[3,4], William T. Zempsky, MD[5,6], Samuel A. Ball PhD[2], and Frank D. Buono, PhD[2]

[1] Department of Epidemiology of Microbial Diseases, Yale School of Public Health, New Haven, CT USA
[2] Department of Psychiatry, Yale School of Medicine, New Haven, CT USA
[3] Child Health Evaluative Sciences, The Hospital for Sick Children, Toronto, Ontario
[4] Institute of Health Policy, Management & Evaluation, University of Toronto, Toronto, Ontario
[5] Department of Pediatrics and Nursing, University of Connecticut, Hartford, CT, USA
[6] Division of Pain and Palliative Medicine, Connecticut Children's Medical Center, Hartford, CT, USA

Corresponding Author:
Lauretta E. Grau, PhD
Yale School of Public Health
PO Box 208034
New Haven, CT 06520
E: lauretta.grau@yale.edu

Key words: COMPLEMENTARY MEDICINE, Adult Neurology, Neurological pain, Neurogenetics, PAIN MANAGEMENT, QUALITATIVE RESEARCH

Word count: 3873; abstract 272

ABSTRACT

Objectives: Neurofibromatosis Type 1 (NF1) is a genetic disorder in which chronic pain commonly occurs. The study sought to understand the needs of individuals with NF1 and pain management experts when adapting a pain self-management mobile health application for individuals with NF1. **Design:** We conducted a series of online, audio-recorded focus groups that were then thematically analyzed. **Setting:** Online focus groups with adults with NF1 currently residing in the U.S. **Participants:** Two types of participants were included: Individuals with NF1 (n = 32 across six focus groups) and pain management experts (n = 10 across three focus groups). **Results:** Six themes across two levels were identified. The Individual level included Lifestyle, Reasons for Using the Mobile App, and Concerns regarding its Use. The Application level included Desired Content, Desired Features, and Format Considerations. Findings included recommendations to grant free access to the application and include a community support feature for individuals to relate and validate one another's experience with pain from NF1. In addition, participants noted the importance of providing clear instructions on navigating the app, the use of an upbeat, hopeful tone, and appropriate visuals.

Conclusions: Both participant groups endorsed the use of iCanCope as an NF1 pain self-management mobile application. Differences between groups was noted regarding However, the NF1 group appeared interested in detailed and nuanced pain tracking capabilities the expert group prioritized tracking information such as mood, nutrition, and activity to identify potential associations with pain. In tailoring the existing iCanCope application for individuals with NF1, attention should be paid to creating a community

support group feature and to tailoring content, features, and format to potential users' specific needs.

Strengths and limitations of this study

- Achieved data saturation, ensuring a deep understanding of the stated research question.
- Online focus group format permitted a time- and cost-efficient opportunity to enable wide geographic representation among participants.
- Unverified NF1 status and low representation among members of minority communities may have limited the credibility and transferability of the findings.
- On-line focus groups limit the ability to observe in-person dynamics or subtle reactions to the various discussion topics.

INTRODUCTION

Neurofibromatosis Type 1 (NF1) is an autosomal dominant genetic disorder affecting an estimated 1 in 3000 individuals worldwide.[1] Common presentations of NF1 can include plexiform neurofibroma tumors, café au lait macules,[2, 3] and extensive dermal neurofibromas.[4] Over 50% of individuals with NF1 report significant pain and discomfort,[5] often stemming from the plexiform neurofibroma tumors.[6] In addition, frequent headaches, orthopedic, and other tumor-related pain[4] may also impair NF1 patients' quality of life.[7]

Current treatment recommendations for chronic pain related to NF1 are limited to pharmacological and surgical methods due to the complexity of the tumors.[8] It is common for individuals with NF1 to employ complementary pain management approaches (e.g., yoga, physical therapy, massage therapy) to manage their pain symptoms.[9] However, there are also challenges to receiving treatment for NF1-related chronic pain such as healthcare system barriers and limited access to self-management techniques,[10] creating a need for innovative, effective, and accessible treatment options.

Mobile health applications, with over 325,000 currently available, have become increasingly popular,[11] including some for self-management of many chronic illnesses.[12, 13] A systematic review concluded that mobile health interventions improved clinical outcomes across varying chronic diseases and increased access to pain management resources at low cost,[14] but once established, they can also be tailored to specific illnesses and conditions.

iCanCope (iCC) is an innovative mobile application originally developed to address pain self-management needs of adolescents and young adults with chronic pain.[15, 16]

The iCC platform was created through a user-centered design approach that involved end-users at every stage and is founded on the principles of cognitive behavioral therapy and the use of mind-body approaches. The core iCC features include daily symptom-tracking, personalized goal-setting to improve pain and functioning, and a toolbox of disease education and pain self-management strategies. A pilot study of iCC demonstrated feasibility of deploying the app among adolescents with chronic pain,[17] and has since been expanded to support pain self-management in youth and young adults with juvenile idiopathic arthritis,[18] sickle cell disease,[19] and post-operative pain, [20] with each adaptation reporting clinically meaningful reductions in pain intensity.

The use of mobile technology to address NF1 patients' needs is a promising strategy to expand access to pain self-management interventions. We therefore proposed to adapt the iCC platform. As a first step, we sought recommendations about the desired features and content that adults with NF1 and pain management experts considered important to include when adapting the iCC mobile health application as well as the perceived acceptability and barriers/facilitators to its use. We reasoned that the multiple perspectives of these individuals would yield a richer and more nuanced assessment of the desired features and content to include in the adaptation.

METHODS

Study Sample

The study involved two participant groups: Adults with NF1 (n = 32) and experts in the treatment and management of NF1-associated pain (n = 10). Inclusion criteria for the NF1 group were: (1) age 18-45 years with an NF1 diagnosis; (2) pain severity

aggregate score $\geq$3 on the Brief Pain Inventory-Short Form (BPI-SF)[21, 22] for pain experienced in the previous two weeks; (3) being proficient in English at the 5th grade level and competent to provide informed consent; (4) agreeing to audio-recording of the focus group session; and (5) residing in the U.S. The exclusion criteria consisted of: (1) moderate to severe cognitive deficits or (2) evidence of moderate to severe depression or anxiety, as assessed by the Patient Health Questionnaire (PHQ)[23] SCID-self-report (SCID-SR)[24] and Generalized Anxiety Disorder scale (GAD-7).[25] Inclusion criteria for the Expert group were: (1) providing healthcare to patients with NF1 (e.g., physicians, nurse practitioners, and psychologists) and (2) agreeing to audio-recording of the focus group session.

Patient and Public Involvement

Patients or the public were not involved in the design, or conduct, or reporting, or dissemination plans of this study.

Recruitment Procedures

Recruitment for the NF1 group consisted of posting an announcement briefly describing the study on the Facebook pages for two NF advocacy groups (NF Northeast and NF Network) and emailing all members of both group registries. Membership in both registries requires self-report of having received an NF diagnosis. Individuals interested in participating were directed to an online survey (www.qualtrics.com) that collected demographic and contact information as well as responses to the BPI, PHQ, GAD-7, and SCID-SR. The research team reviewed the results of these clinical

measures, and individuals meeting the inclusion requirements then received a list of focus group discussion (FGD) dates/times from which to select. Recruitment for the Expert group occurred via Website postings about the study on the US Association for the Study of Pain and the NF Network clinician page. Prior to beginning each FGD, participants were provided with a brief description of the study and informed of the voluntary nature of their participation. All participants provided verbal informed consent prior to participating in the FGD session.

After completing the FGD, participants in the NF1 and expert groups received a $25 and $100 gift card, respectively. The Institutional Review Board at Yale University approved all study materials and waived the need to obtain written consent.

Data Collection and Analysis

Each FGD session lasted approximately one hour and occurred via a secure, password-enforced video conference (www.zoom.com). Two members of the research team (FB, KL) with prior training in conducting focus groups jointly facilitated the sessions.

An interview guide was developed specifically for this study (see Supplementary Material). Only the NF1 group sessions began by asking whether participants owned smartphones. All participants were then asked their opinions about using a phone-based application to address NF1 pain management. A brief presentation then occurred that described the existing iCC mobile application and included four screen shots from the application. The remaining session time was guided by that description, the four screen shots, and the four domains included in the FGD guide: (1) perceived barriers and facilitators to using a mobile application to manage pain; (2) perceived acceptability and benefits of the proposed application; (3) contexts within which the application would most likely be used; and (4) recommendations for possible additions or modifications to the application. Demographic information for the NF1 group included age and gender, collected at eligibility screening, and for the Expert group included professional training/title, years in practice, and gender, collected at the beginning of each FGD. The audio-recorded transcripts were professionally transcribed verbatim.

An existing codebook from a prior iCC study[26] was modified for use in the current study. Members of the data analysis team (FB, KL, LG, JS, CL) met regularly to review transcripts and discuss codebook development. Interviews continued until data saturation had been achieved.[27, 28] The final codebook included 29 codes, three of which were new codes from the Expert group sessions. Acceptable inter-coder agreement was established after independently coding two transcripts. The remaining transcripts were then independently coded by both coders (FB, KL). The coding team continued to meet periodically to resolve coding discrepancies (through consensus), address coding questions, and further refine code definitions as needed. The coded transcripts were then entered into Dedoose (www.dedoose.com, version 8.3.47) for analysis. The coding and analysis process continued in an iterative fashion using thematic analysis.[29, 30] Negative instances where the data did not fit the existing themes were identified as part of the confirmability process.[31] Descriptive statistics were calculated for the demographic data. The entire research team reviewed all analytic findings.

RESULTS

A total of nine FGDs—six NF1 groups (n = 32) and three pain management expert groups (n = 10)—occurred between January and May 2020. Previous research has shown intrinsic motivation[32] to be the critical, first step in promoting future goal-oriented behaviors.[33] We therefore sought to identify themes that shaped motivation to use the proposed mobile application and generated a total of six themes across two levels (i.e., Individual and Application) as seen in the conceptual model in Figure 1. The themes of Lifestyle, Reasons for Using the Mobile App, and Concerns regarding Use of the Mobile Application were identified at the Individual Level. The App Level included the themes of Desired Content, Desired Features, and Format Considerations.

(Insert Figure here)

Description of the Study Sample

A total of 42 individuals participated in the study (Table 1). Overall, the NF1 group was generally young and predominantly female. Except for one participant, all owned smartphones. Most within the NF1 group were experiencing moderate pain at screening. The Expert group consisted entirely of individuals with post-graduate training in their respective fields and had, on average, over 19 years of professional experience. Similar to the NF1 group, the majority of expert participants were female.

Table 1. Characteristics of the Study Sample (N = 42)

	NF1 Group (n = 32)	Expert Group (n = 10)
Age (years)[1]	31.2 (4.9)	—
Range	18 - 39	
Gender[2]		
Male	6 (19%)	4 (40%)
Female	25 (78%)	6 (60%)
Other	1 (4%)	—
Aggregate Pain Severity[1,3]	4.8 (1.7)	—

Aggregate Pain Interference[1,4]	4.9 (2.6)	—
Professional Training/Title[2]		
Pain Psychologist	—	2 (20%)
Neurologist	—	5 (50%)
Primary Care Physician	—	2 (20%)
Neuro-oncologist	—	1 (10%)
Years in Practice[1]	—	19.2 (7.2)
Range		7 - 38

[1] Mean (S.D.)
[2] N (%)
[3] BPI-SF; 4 items for pain severity, possible range of scores is 0 - 10
[4] BPI-SF, 7 items for pain interference, possible range of scores is 0 – 10

Individual Level Themes

The Individual Level focused on the life situations of individuals with NF1 that were thought to influence motivation to use the proposed mobile application.

Table 2. Individual Level Themes and Quotes

Theme 1: Lifestyle	
Affordability/Accessibility sub-theme	Where I work there's certain applications that are blocked so it would be really a pain for lack of a better term for me to have to kind of remember everything and write it down later. (Participant 26, NF1 group)
	Unfortunately, in rural areas such as southwest Pennsylvania where I live or West Virginia a lot of people just don't have connections that are that strong sometimes. (Participant 24, NF1 group)
	There's always a lot of barriers in terms of access, economics, et cetera. (Participant 103, Expert group)
Time sub-theme	Sometimes I get up late and it's just something that I need to do every day but tend to forget if I'm hustling out the door or something. (Participant 7, NF1 group)
	I'm very bad about inputting stuff and keeping diaries and stuff because I have two toddlers at home, so that takes a lot of my time, too. (Participant 17, NF1 group)
	Other barriers are typically time… (Participant 106, Expert group)
Theme 2: Reasons for Using the Mobile Application	
Engagement sub-theme	I like apps that give me badges and awards for accomplishing my goals. (Participant 10, NF1 group)
	There has to be a way that they can, early in their treatment, start to see positive change, positive success

	so they get these little wins in the beginning and that's just the psychology of motivation and how you sort of really start to get that ball rolling. (Participant 104, Expert group)
Hope sub-theme	They have to be able to hold hope that they have the opportunity to be efficacious in improving the situation. (Participant 104, Expert group)
Theme 3: Concerns about Using the App	Who could potentially access that information? Could health insurance companies access…I'm worried about who has access to that and how they could potentially use it. (Participant 14, NF1 group)
	[Use of stars or points] can be very effective if they're tooled in the right way…but they [users] have to feel like that the goals are attainable. (Participant 104, female, Expert group)

Theme 1: Lifestyle (Individual Level)

Two sub-themes were identified within the Lifestyle theme (Table 2): Affordability/Accessibility and Time. The former focused on financial or other considerations that could influence ease of access. Both groups recognized that the mobile application needed to be "very cheap" or free and not consume much data (i.e., bottlenecking). Access issues related to geographic location or application use that depended upon external factors such as Internet signal strength or workplace access policies were also mentioned but were beyond what could be addressed within the application.

Time, the second sub-theme, focused on the busy lifestyles of most NF1 group members, usually involving parenting or work responsibilities. Both participant groups recognized that use of the application needed to easily fit into users' schedules. Barriers to its use included forgetting to access the application during busy times or when confronted with unexpected outside demands.

Theme 2: Reasons for Using the Mobile App (Individual Level)

This theme concerned potential factors thought to directly increase motivation to use the mobile application, primarily through rapid user engagement with program content and activities. It included two sub-themes: Engagement and Hope (Table 2).

The Engagement sub-theme focused on the fact that users would be more likely to use the application if they achieved improvements or successes early on. As one expert noted, “The ultimate aim is showing progress.” Both participant groups also believed that earning rewards (e.g., badges, points) can promote engagement with and continued use of the application. Participants often referred to this as “buy-in” and viewed it as critical to program uptake.

Similarly, instilling a sense of hope was seen as essential to promoting engagement and continued use of the program. Instilling hope was not considered to be limited to explicit messages of hope but could also result from individuals with NF1 recognizing improvements in function, quality of life, or having their NF1 pain validated by others.

Theme 3: Concerns regarding Use of the Mobile App (Individual Level)

The NF1 group seemed particularly concerned about privacy and access. They worried about who would have access to their health data and whether access to such data could jeopardize health insurance coverage or pose an unforeseen risk (Table 2). As previously mentioned in the Affordability/Accessibility sub-theme, participants were also concerned that Internet coverage or work restrictions could limit use of the application. Members of the Expert group were concerned that if goals were not realistic and achievable within an appropriate timeframe, some users might discontinue using the mobile application.

The Application Level

The three themes included at the Application Level focused on elements in the proposed mobile application that could increase users' motivation to use the product. The themes primarily concerned the content to be included and the functionality and features of the proposed product.

Theme 4: Desired Content (App Level)

Participants identified the important types of information about chronic pain and its management that is needed by individuals with NF1. It included the sub-themes of "Pain 101," Self-care, and Clinical Care (Table 3).

Table 3. Application Level Themes and Quotes for Desired Content and Desired Features

Theme 4: Desired Content	
"Pain 101" sub-theme	I do think the pain education piece and how pain works in the brain would be really great for NF patients. (Participant 101, Expert group)
	…how to work through and navigate the healthcare system, how to talk to healthcare providers about their pain and options. (Participant 106, Expert group)
	It's the specific exercises…To have it be specific, broken down like a menu. Headaches? Do this. (Participant 2, NF1 group)
Self-care sub-theme	I think the mindfulness or meditation, yoga, breathing techniques would be extremely helpful. (Participant 26, NF1 group)
	…just getting them exposed to different complementary and alternative therapies has value, so even if you don't teach it in the app… (Participant 108, Expert group)
Clinical Care sub-theme	I did a study for NF with ACT, that's Acceptance and Commitment Treatment. I liked it because I think if you were going to do something with CBT [cognitive-behavior therapy], if you could incorporate some ACT as well, that would be great because to help you rethink about your pain might be… (Participant 10, NF1 group)
	…evidence-based treatment for chronic pain involves a whole host of cognitive and behavioral strategies including pacing for pain and mindfulness, relaxation, catching catastrophic thoughts about pain and about the future, as well as lifestyle changes like nutrition and sleep hygiene. (Participant 101, Expert group)
Theme 5: Desired Features	

Tracking Capabilities sub-theme	I had to go through boxes of medical records to figure out and remember all the different doctors…so if there was a way to store some information… (Participant 3, NF1 group)
	I think one cool thing would be if there was an outline of a body that you could blow up and click on the part exactly that hurts and kind of color it in or shade it in or something” (Participant 19, NF1 group)
	But if here’s a graph then he or she also says here’s the pain I was having and here’s what else what was going on at that time and what I think may have contributed, then I may be able to offer some further intervention. (Participant 109, Expert group)
	I think tracking mood is hugely important. We know that anxiety and depression very commonly and understandably co-occur with chronic pain and medical conditions. We also know that mood and stress and anxiety and depression impact and amplify pain. (Participant 101, Expert group)
Community sub-theme	…things where we could actually keep up with one another, talk to one another, just get things off our chest. I think that would be incredibly beneficial. (Participant 6, NF1 group)
	I think having positive or some opportunities to connect with other people that really understand the disease entity can be helpful too, as long as they’re all working toward making gains and not just being really steeped and sad about the disease process. (Participant 104, Expert group)
	It was years before I ever met anybody else with NF and it’s nice to know that there are other people out there…it would be nice on this app to be able to have maybe a comment section so that people can say, yup, I was there, or this is how I cope. (Participant 14, NF1 group)
	…people share incredible stuff on [message boards], so sharing pain may be nothing compared to sharing their personal stories… (Participant 107, Expert group)
External Resources sub-theme	I think of it as a way to maybe link certain maybe research articles or advice [that]…afterwards if you log a specific instance of pain and there’s something that could pop up that you could read… (Participant 6, NF1 group)
	We use [name of app] …And it’s been really helpful for things like CRPS [complex regional pain syndrome], potentially amputation pain, and so it’s part of cognitive retraining for the disorganized cognitive input from a missing limb or a dysfunctional limb. (Participant 105, Expert group)
Future Features sub-theme	One thing I’d like to see for the physical activity, if it could import data from say [names of activity apps] or whatever. If you use one of those apps, it would be one less thing for the user to do. (Participant 20, NF1 group)
	I want my app to be able to function at least as good as it can with no Internet connection and automatically update the database when I get connection. (Participant 24, NF1 group)
	One of the apps I used to use was for water and as you drank water it grew a plant. Well, maybe something interactive based on the age ranges of who might use it. (Participant 9, NF1 group)
	…if [patients] can print out a graph or somehow send the graphical representation that we’ve like scanned that into the medical record, that’s been helpful (Participant 108, Expert group)
	I think one really important feature that a lot of other applications that I use lack is integration with MyChart and other things that my

	doctors would have access to so that everybody can just see it rather than having to go back and forth with all the different doctors (Participant 26, NF1 group)

The experts believed that accurate knowledge about the topic of pain was important in motivating individuals to use the mobile application— “a very essential piece of this work, not sufficient, but necessary” and referred to it as “Pain 101” information. This included information on the physiological basis of pain, neuropathic and nociplastic pain, the difference between acute and chronic pain, and NF1-specific pain. They also noted that users should receive instruction about how to successfully navigate the healthcare system and effectively interact with their healthcare providers.

By contrast, the NF1 group appeared most interested in learning about symptom recognition, recommended analgesics, and strategies for targeting specific types of pain.

The Self-care sub-theme dealt with pain treatment approaches potential users could learn and then do independently or in group settings. Both participant groups offered many suggestions about possible pain mitigation strategies. These could be divided into two types of self-care strategies: physical and psychobehavioral. The former, endorsed by both groups, included movement-based activities such as martial arts, yoga or “grounding exercises,” and pacing. Only the Expert group suggested improvement of sleep hygiene and use of heat and ice as physical approaches to self-management of NF1 pain. Both groups suggested psychobehavioral approaches (i.e., those requiring only brief training with a qualified professional) such as deep, circular, or meditational breathing and music or aroma therapy.

The final sub-theme, Clinical Care, focused on pain treatments that required meeting with a trained professional for longer periods than typically required in Self-care approaches. Yet, similar to the Self-care sub-theme, the treatments could be divided into physical and psychobehavioral approaches. Only the expert group identified physical approaches such as physical therapy or nutrition care. Longer term psychobehavioral approaches included cognitive behavior therapy, acceptance and commitment therapy, guided imagery, biofeedback, and mindfulness training.

Theme 5: Desired Features (App Level)

The Desired Features theme included four sub-themes: (1) Tracking capabilities, (2) Community, (3) External Resources, and (4) Future Features. This theme focused on the functionality of the mobile application and the desire for links to external resources (Table 3).

Tracking of various events and information was considered by both participant groups to be an important feature of the mobile application. It included tracking and monitoring pain level, mood, activity, sleep, and stress, all considered to be important potential pain triggers. Both groups also believed that maintaining notes or a diary was important but for somewhat different reasons. The NF1 group viewed this feature as a potential memory aid that could enable patients to “talk about [pain symptoms] more intelligently to a provider.” It could serve as a reminder for exercising, logging information in the application, or to track meditation and physical activity. The NF1 group also expressed interest in recording their surgical and medication histories and having the capability of tracking pain details such as onset, location, quality, and level of pain.

By contrast, the Expert group appeared to be interested in a less detailed approach to tracking pain, albeit sufficiently detailed to be able to identify associations between pain and such things as mood, activity, or treatment efficacy.

Both participant groups were enthusiastic about creating an online community that could serve as a resource for socioemotional support and NF1-relevant information (e.g., local events, providers). Members of the Expert group cautioned that it should not be a venue for complaining about having the disease, however. The NF1 group offered many suggestions about the format (e.g., group size, age restrictions, creation of a buddy system), although none emerged as the preferred format. Both groups also agreed that sharing experiences could help normalize having NF1 and help individuals feel less isolated.

Participants believed that external resources such as links to other mobile applications could be helpful to NF1 patients. The NF1 group was interested in links to relevant articles and treatment guidelines. The Expert group suggested links that could assist individuals with NF1 in self-management of their disease, including relaxation and pain sites.

Both groups identified some features that were currently not feasible to include in the mobile application, as they would require extensive resources, negotiations with other applications, or lengthy modifications to the current iCC. They can be considered for future modifications to the application. Suggestions included automatic data upload at the next sign-in to make it more convenient for users whose Internet access is blocked at work or in certain geographical locations, printable reports, 24/7 access to medical advice, and a visual graphic representation of progress toward a specific goal.

Other suggestions, primarily from the NF1 group, were currently unrealistic to implement because they require formalizing agreements with healthcare institutions. For example, they mentioned wanting the capability to automatically share information with healthcare providers or access existing x-rays when at another location.

Theme 6: Format Considerations (App Level)

The final theme included two sub-themes (Table 4): User Interface and Presentation/Tone. The first addressed issues that would increase users' ease of use, and the second focused on the visual appeal and tone of the content presented.

Table 4. Application Level Theme and Quotes for Format Considerations

Theme 6: Format Considerations	
User Interface sub-theme	I need consistency. Kind of like if you're going to do updates and stuff like that, it needs to not be completely reconfigured. (Participant 1, NF1 group)
	If I can add in my own entries along with that, that would motivate me to use it more. (Participant 18, NF1 group)
	…then you want a very simple front page and then links or tabs. (Participant 105, Expert group)
Presentation/Tone sub-theme	I think having some visuals as well would be useful. Yeah, visual, not horrid looking. (Participant 1, NF1 group)
	I would want it to be simple and fun and use language that I use, not language a doctor would use. I can look up words, but sometimes I don't want to. (Participant 4, NF1 group)
	I think considering color is important…We didn't really talk about that but, the whole visual appeal. Because if it's not appealing or if it's going to make a person feel worse then that's counter, then that's going against what [the goal of the application is]. (Participant 102, Expert group)
	When I was looking at your app, it made me think about music and how that can be useful for people's state of mind, which you can't separate the body from the mind so I think that would be great in chronic pain. (Participant 103, Expert group)

Both groups understood the importance of User Interface, that motivation to use the mobile application could be heavily influenced by its ease of use. The Expert group suggested having explicit instructions on how to navigate and use the application. Participants from both groups recommended tailoring the application to users' specific preferences on pain-tracking measures and format (e.g., numeric vs. figural, pain descriptors) and including an "Other" option so users could further qualify their responses to forced-choice questions. They also identified features to enhance time-efficiency such as auto-population of key information, use of drop-down menus, and consistency in format and content when updating.

Critical elements in motivating individuals to use the mobile application were Tone and Presentation, the final sub-theme. Participants described the ideal application as being "approachable and digestible" and "simple and fun." Visual appeal was important, noting that the font and use of color could influence use. Careful selection of NF1 photographs and language could also be used to set an appropriate tone. Members of the NF1 group believed that daily tips or inspiring quotes could enhance engagement, and experts recommended varying the messages to keep the content fresh. Both groups agreed that music could help set a positive tone as well.

DISCUSSION

Few studies have examined the perceived benefits, desired content and features, or efficacy of mobile health applications on coping with NF1-related chronic pain. To our knowledge, the current study is the only one to examine some of these issues from the perspectives of the targeted end-users and pain management experts. We believe that this is essential information to ensure that the product is scientifically accurate and relevant while also satisfying targeted end-users' needs and preferences.

Both groups were enthusiastic about the prospect of adapting the iCC mobile application for individuals with NF1 with chronic pain. They believed that tailoring the program to accommodate users' lifestyle needs and affordability, incentivizing its use, fostering an NF1 community support group, and using appropriate and acceptable tone and format features will encourage uptake and continued use of the mobile application. In addition, offering NF1 and coping skills information and an online support community in a single mobile application renders it both convenient and comprehensive in scope.

Participants believed that flexibility in fitting into potential users' current situation was important. They noted that the application should be affordable, easy to use, and convenient; it should fit users' schedules, efficiently capture the desired data, and be modifiable as users' life situations change. These findings support previous research on features needed to ensure uptake and ease of use of the mobile application[34, 35] as well as health-related mobile application users' privacy and security concerns.[36, 37]

With respect to the issue of promoting sustained use of the application, participants from both groups noted the importance of including realistic and achievable goals. Early improvements might involve pain reduction or changes in pain attitudes that could improve mood, function, or quality of life. Consistent with the notion of contingency management,[38, 39] participants identified strategies involving points, levels, prizes, or well-established reinforcement schedules to incentivize continued use. These findings suggest the importance of considering contingency management when developing an application to promote health behaviors.[40,41]

Regarding the pain symptom tracking feature, the two groups had somewhat different views on what type of pain data are most desired. The NF1 group appeared interested in gathering as detailed and nuanced pain information as possible. By contrast, the Expert group appeared more interested in tracking information that might identify potential associations between pain, mood, nutrition, activity, and stress levels; they believed that noting these patterns would be more useful than tracking pain intensity, duration, quality, and directionality. Consistent with previous research,[42] the two participant groups expressed different perspectives on and desires for tracking chronic pain. The data suggest that to accommodate these disparate views, the pain tracking feature should permit simultaneous tracking of several constructs (e.g., pain, mood, activity) while also providing a flexible format so that patients can select the level of detail and pain metrics they prefer. One cautionary note, however, would be to avoid the potential for users to become fixated on their pain symptoms to the detriment of recognizing the associations between their pain and the other constructs. Based on input from our Expert group, we plan to develop an app that is engaging without over-focusing on tracking or complaining about symptoms and subsequently assess its effectiveness and potential positive/negative effects.

Community support, particularly for individuals with rare diseases such as NF1, can be an important coping resource.[43,44] Previous research on online community support platforms for individuals with rare diseases has noted that they are used for both informational and socioemotional support purposes.[45, 46] The value of community support has been identified within the context of cultural and racial socialization,[47, 48] and the study corroborates these findings. Both groups noted how the creation of an online NF1 community would provide users the opportunity to share and thereby validate their experiences with the disease and chronic pain, provide socioemotional encouragement, and potentially motivate continued use of the mobile application.

The study supported previous findings about visual appeal, tone, ease of use, instructions on effectively navigating app options, tailoring mHealth interventions to personal needs and preferences, and the use of appropriate visuals.[49, 50] This last feature is particularly important with a potentially disfiguring disease such as NF1. A novel finding from this study, however, is the importance of establishing an upbeat tone that instills hope.

We note several study limitations. First, the findings may have limited transferability, given the modest racial and ethnic diversity of the study sample. Second, lack of external confirmation of self-reported NF1 diagnoses may have limited the credibility of the results. Third, the myriad suggestions proffered may be attributed to social desirability wherein participants sought to identify as many options as possible in order to “please” the group facilitators. Nonetheless, these options made possible a deeper analysis of potential format and content considerations when adapting the iCC application. Finally, virtual FGDs limit the ability to observe subtle, in-person dynamics during discussions. We chose this format, however, prior to the onset of the COVID-19 pandemic, based upon our desire to collect data in a time- and cost-efficient manner across diverse geographic regions and living situations.

Conclusions

The study findings suggest that adapting the iCC mobile application for use in the self-management of NF1-associated chronic pain was seen by adults with NF1 and chronic pain experts as a welcomed addition to the armamentarium of NF1 treatment resources. Careful attention should be paid to the creation of a community support group option as well as program content, format, and ability to tailor it to users' specific needs.

Acknowledgements: We are indebted to Karen Peluso, Director of NF Northeast and Kim Bischoff, Director of NF Network for their help and commitment. We also thank the study participants for sharing with us their time and insights.

Funding: This work was supported by the U.S. Army Medical Research Materiel Command endorsed by the U.S. Army, through the Congressionally Directed Medical Research Programs' Neurofibromatosis Research Program under Award No. W81XWH-19-1-0618.

Competing Interests Statement: None declared.

Author Contributions:

LG- Manuscript preparation and review, study design and data analysis, development of interview guide

KL- Daily oversight of project, focus group facilitation, data management and analysis, manuscript preparation and review

CL- Development of interview guide, manuscript preparation and review

JS- Development of interview guide, manuscript preparation and review

WZ- Supervision of the project, manuscript review

SB- Supervision of the project, manuscript review

FB- Study design and data analysis, led the focus groups, development of interview guide, manuscript preparation and review, oversight for the general conduct of this study

Data Sharing Statement: Requests to receive the qualitative data should be addressed to the Corresponding Author. Upon review of the request, the data may be made available.

Ethics Statement: The Human Subjects Committee of the Yale Human Research Protection Program of Institutional Review Boards at Yale University approved Protocol #2000025437.

Figure Legend: Six themes identified within the data were thought to shape motivation to use the mobile application. Three could be attributed to personal characteristics of the targeted end-users and were thought to directly influence motivation. The remaining three focused on content and qualities of the application itself and were thought to indirectly influence motivation to use the application.

REFERENCES

1. Anderson JL, Gutmann DH. Neurofibromatosis type 1. *Handb Clin Neurol* 2015;132:75-86. PMID: 26564071. doi: 10.1016/B978-0-444-62702-5.00004-4.
2. Kodra Y, Giustini S, Divona L, Porciello R, Calvieri S, Wolkenstein P, et al. Health-related quality of life in patients with neurofibromatosis type 1. A survey of 129 Italian patients. *Dermatology* 2009;218:215-20. PMID: 19088462. doi: 10.1159/000187594.
3. Radtke HB, Sebold CD, Allison C, Haidle JL, Schneider G. Neurofibromatosis type 1 in genetic counseling practice: recommendations of the National Society of Genetic Counselors. *J Genet Couns* 2007;16:387-407. PMID: 17636453. doi: 10.1007/s10897-007-9101-8.
4. Tonsgard JH. Clinical manifestations and management of neurofibromatosis type 1. *Semin Pediatr Neurol* 2006;13:2-7. PMID: 16818170. doi: 10.1016/j.spen.2006.01.005.
5. Brems H, Beert E, de Ravel T, Legius E. Mechanisms in the pathogenesis of malignant tumours in neurofibromatosis type 1. *Lancet Oncol* 2009;10:508-15. PMID: 19410195. doi: 10.1016/s1470-2045(09)70033-6.
6. North KN, Riccardi V, Samango-Sprouse C, Ferner R, Moore B, Legius E, et al. Cognitive function and academic performance in neurofibromatosis. 1: consensus statement from the NF1 Cognitive Disorders Task Force. *Neurology* 1997;48:1121-7. PMID: 9109916. doi: 10.1212/wnl.48.4.1121.
7. Vranceanu A-M, Merker VL, Park E, Plotkin SR. Quality of life among adult patients with neurofibromatosis 1, neurofibromatosis 2 and schwannomatosis: a systematic review of the literature. *J Neurooncol* 2013;114:257-62. doi: 10.1007/s11060-013-1195-2.
8. Sabatini C, Milani D, Menni F, Tadini G, Esposito S. Treatment of neurofibromatosis type 1. *Curr Treat Options Neurol* 2015;17(6):355. PMID: 25917340. doi: 10.1007/s11940-015-0355-4.
9. Buono FD, Grau LE, Sprong ME, Morford KL, Johnson KJ, Gutmann DH. Pain symptomology, functional impact, and treatment of people with Neurofibromatosis type 1. J *Pain Res* 2019;12:2555-61. PMID: 31692483. doi: 10.2147/jpr.S209540.
10. Martin S, Wolters PL, Toledo-Tamula MA, Schmitt SN, Baldwin A, Starosta A, et al. Acceptance and commitment therapy in youth with neurofibromatosis type 1 (NF1) and chronic pain and their parents: A pilot study of feasibility and preliminary efficacy. *Am J Med Genet A* 2016;170:1462-70. PMID: 27021207. doi: 10.1002/ajmg.a.37623.
11. Ferrara G, Kim J, Lin S, Hua J, Seto E. A Focused Review of Smartphone Diet-Tracking Apps: Usability, Functionality, Coherence With Behavior Change Theory, and Comparative Validity of Nutrient Intake and Energy Estimates. *JMIR Mhealth Uhealth* 2019;7:e9232. PMID: 31102369. doi: 10.2196/mhealth.9232.
12. Birkhoff SD, Smeltzer SC. Perceptions of Smartphone User-Centered Mobile Health Tracking Apps Across Various Chronic Illness Populations: An Integrative

Review. *J Nurs Scholarsh* 2017;49:371-8. PMID: 28605151. doi: 10.1111/jnu.12298.

13. Sarkar U, Gourley GI, Lyles CR, Tieu L, Clarity C, Newmark L, et al. Usability of Commercially Available Mobile Applications for Diverse Patients. *J Gen Intern Med* 2016;31:1417-26. PMID: 27418347. doi: 10.1007/s11606-016-3771-6.
14. Whitehead L, Seaton P. The Effectiveness of Self-Management Mobile Phone and Tablet Apps in Long-term Condition Management: A Systematic Review. *J Med Internet Res* 2016;18:e97. PMID: 27185295. doi: 10.2196/jmir.4883.
15. Stinson JN, Lalloo C, Harris L, Isaac L, Campbell F, Brown S, et al. iCanCope with Pain™: User-centred design of a web- and mobile-based self-management program for youth with chronic pain based on identified health care needs. *Pain Res Manag* 2014;19:257-65. PMID: 25000507. doi: 10.1155/2014/935278.
16. Stinson JN, White M, Isaac L, Campbell F, Brown S, Ruskin D, et al. Understanding the information and service needs of young adults with chronic pain: perspectives of young adults and their providers. *Clin J Pain* 2013;29:600-12. PMID: 23328333. doi: 10.1097/AJP.0b013e31826dce65.
17. Lalloo C, Hundert A, Harris L, Pham Q, Campbell F, Chorney J, et al. Capturing Daily Disease Experiences of Adolescents With Chronic Pain: mHealth-Mediated Symptom Tracking. *JMIR Mhealth Uhealth* 2019;7:e11838. PMID: 30664472. doi: 10.2196/11838.
18. Lalloo C, Harris LR, Hundert AS, Berard R, Cafazzo J, Connelly M, et al. The iCanCope pain self-management application for adolescents with juvenile idiopathic arthritis: a pilot randomized controlled trial. *Rheumatology (Oxford)* 2021;60:196-206. PMID: 32613229. doi: 10.1093/rheumatology/keaa178.
19. Palermo TM, Zempsky WT, Dampier CD, Lalloo C, Hundert AS, Murphy LK, et al. iCanCope with Sickle Cell Pain: Design of a randomized controlled trial of a smartphone and web-based pain self-management program for youth with sickle cell disease. *Contemp Clin Trials* 2018;74:88-96. PMID: 30316999. doi: 10.1016/j.cct.2018.10.006.
20. Birnie KA, Campbell F, Nguyen C, Lalloo C, Tsimicalis A, Matava C, et al. iCanCope PostOp: User-Centered Design of a Smartphone-Based App for Self-Management of Postoperative Pain in Children and Adolescents. *JMIR Form Res* 2019;3:e12028. PMID: 31008704. doi: 10.2196/12028.
21. Cleeland CS, Ryan KM. Pain assessment: global use of the Brief Pain Inventory. *Ann Acad Med Singap* 1994;23:129-38. PMID: 8080219.
22. Palos GR, Mendoza TR, Mobley GM, Cantor SB, Cleeland CS. Asking the community about cutpoints used to describe mild, moderate, and severe pain. *J Pain* 2006;7:49-56.
23. Kroenke K, Spitzer RL, Williams JB. The PHQ-9: validity of a brief depression severity measure. *J Gen Intern Med* 2001;16:606-13. PMID: 11556941. doi: 10.1046/j.1525-1497.2001.016009606.x.

24. Brodey B, Purcell SE, Rhea K, Maier P, First M, Zweede L, et al. Rapid and Accurate Behavioral Health Diagnostic Screening: Initial Validation Study of a Web-Based, Self-Report Tool (the SAGE-SR). *J Med Internet Res* 2018;20:e108. PMID: 29572204. doi: 10.2196/jmir.9428.

25. Spitzer RL, Kroenke K, Williams JB, Lowe B. A brief measure for assessing generalized anxiety disorder: the GAD-7. *Arch Intern Med* 2006;166):1092-7. PMID: 16717171. doi: 10.1001/archinte.166.10.1092.

26. Ahola Kohut S, LeBlanc C, O'Leary K, McPherson AC, McCarthy E, Nguyen C, et al. The internet as a source of support for youth with chronic conditions: A qualitative study. *Child Care Health Dev* 2018;44:212-20. PMID: 29082537. doi: 10.1111/cch.12535.

27. Guest G, Bunce A, Johnson L. How Many Interviews Are Enough?:An Experiment with Data Saturation and Variability. *Field Methods* 2006;18:59-82. doi: 10.1177/1525822x05279903.

28. Hennink MM, Kaiser BN, Marconi VC. Code Saturation Versus Meaning Saturation: How Many Interviews Are Enough? *Qual Health Res* 2017;27:591-608. PMID: 27670770. doi: 10.1177/1049732316665344.

29. Braun V, Clarke V. Using thematic analysis in psychology. Qualitative Research in Psychology. 2006 2006/01/01;3(2):77-101. doi: 10.1191/1478088706qp063oa.

30. Braun V, Clarke V. Successful qualitative research: A practical guide for beginners: sage; 2013. ISBN: 1446289516.

31. Henwood KL, Pidgeon NF. Qualitative research and psychological theorizing. Br J Psychol. 1992 Feb;83 (Pt 1):97-111. PMID: 1559146. doi: 10.1111/j.2044-8295.1992.tb02426.x.

32. Deci EL, Ryan RM. Intrinsic Motivation and Self-Determination in Human Behavior.1985. New York:Springer Science+Business Media. ISBN: 978-1-4899-2273-1.

33. Ajzen I. From Intentions to Actions: A Theory of Planned Behavior. In: Kuhl J, Beckmann J, editors. Action Control: From Cognition to Behavior. Berlin:Springer-Verlag Berlin Heidelberg; 1985. p. 11-39. ISBN: 978-3-642-69746-3

34. O'Brien T, Rosenthal A. Preferred Features in Mobile Health Applications for Kidney Transplant Recipients: A Qualitative Approach. *Nephrol Nurs J* 2020;47:529-36. [published Online First: 2020/12/31]

35. Zapata BC, Fernández-Alemán JL, Idri A, et al. Empirical Studies on Usability of mHealth Apps: A Systematic Literature Review. *J Med Syst* 2015;39:1. doi: 10.1007/s10916-014-0182-2

36. Martínez-Pérez B, de la Torre-Díez I, López-Coronado M. Privacy and security in mobile health apps: a review and recommendations. *J Med Syst* 2015;39:181. doi: 10.1007/s10916-014-0181-3

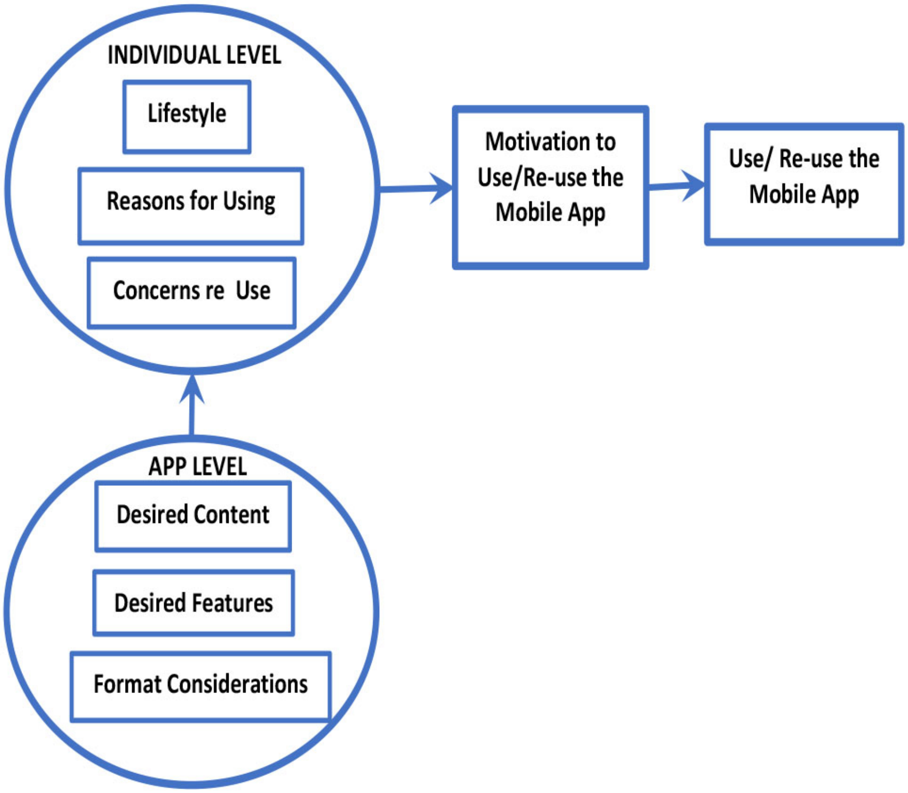
37. Agarwal P, Gordon D, Griffith J, Kithulegoda N, Witteman HO, Sacha Bhatia R, et al. Assessing the quality of mobile applications in chronic disease management: a

scoping review. *NPJ Digit Med* 2021;4:46. PMID: 33692488. doi: 10.1038/s41746-021-00410-x.

38. Higgins ST, Petry NM. Contingency management. Incentives for sobriety. *Alcohol Res Health* 1999;23:122-7. PMID: 10890806.
39. Higgins ST, Silverman K. Motivating behavior change among illicit-drug abusers: Research on contingency management interventions. Washington, DC, US: American Psychological Association; 1999. ISBN: 1-55798-570-7.
40. Máca V, Ščasný M, Zvěřinová I, et al. Incentivizing Commuter Cycling by Financial and Non-Financial Rewards. *Int J Environ Res Public Health* 2020;17:6033 doi: 10.3390/ijerph17176033
41. Carpenter VL, Hertzberg JS, Kirby AC, et al. Multicomponent smoking cessation treatment including mobile contingency management in homeless veterans. *J Clin Psychiatry* 2015;76:959-64. doi: 10.4088/JCP.14m09053
42. Kovačević I, Kogler VM, Turković TM, Dunkić LF, Ivanec Ž, Petek D. Self-care of chronic musculoskeletal pain - experiences and attitudes of patients and health care providers. *BMC Musculoskelet Disord* 2018;19:76. PMID: 29514616. doi: 10.1186/s12891-018-1997-7.
43. Martin S, Struemph KL, Poblete A, et al. An Internet support group for parents of children with neurofibromatosis type 1: a qualitative analysis. *J Community Genet* 2018;9:327-34. doi: 10.1007/s12687-018-0360-x
44. Martin S, Wolters PL, Baldwin A, et al. Attitudes about internet support groups among adolescents and young adults with neurofibromatosis type 1 and their parents. *J Genet Couns* 2014;23:796-804. doi: 10.1007/s10897-014-9688-5
45. Lasker JN, Sogolow ED, Sharim RR. The role of an online community for people with a rare disease: content analysis of messages posted on a primary biliary cirrhosis mailinglist. *J Med Internet Res* 2005;7:e10. PMID: 15829472. doi: 10.2196/jmir.7.1.e10.
46. Doyle M. Peer Support and Mentorship in a US Rare Disease Community: Findings from the Cystinosis in Emerging Adulthood Study. *Patient* 2015;8:65-73. PMID: 25231828. doi: 10.1007/s40271-014-0085-9.
47. Litchman ML, Edelman LS, Donaldson GW. Effect of Diabetes Online Community Engagement on Health Indicators: Cross-Sectional Study. *JMIR Diabetes* 2018;3:e8. doi: 10.2196/diabetes.8603
48. Young SD, Holloway I, Jaganath D, et al. Project HOPE: online social network changes in an HIV prevention randomized controlled trial for African American and Latino men who have sex with men. *Am J Public Health* 2014;104:1707-12. doi: 10.2105/ajph.2014.301992
49. Llorens-Vernet P, Miró J. Standards for Mobile Health-Related Apps: Systematic Review and Development of a Guide. *JMIR Mhealth Uhealth* 2020;8:e13057. PMID: 32130169. doi: 10.2196/13057.
50. Woods L, Duff J, Cummings E, Walker K. Evaluating the Development Processes of Consumer mHealth Interventions for Chronic Condition Self-management: A

Scoping Review. *Comput Inform Nurs* 2019;37:373-85. PMID: 31135468. doi: 10.1097/cin.0000000000000528.

Figure 1 Conceptual Framework and Themes for Motivation to Use the Mobile Application


INDIVIDUAL LEVEL
Lifestyle
Reasons for Using
Concerns re Use
Motivation to Use/Re-use the Mobile App
Use/ Re-use the Mobile App
APP LEVEL
Desired Content
Desired Features
Format Considerations

Legend: Six themes identified within the data were thought to shape motivation to use the mobile application. Three could be attributed to personal characteristics of the targeted end-users and were thought to directly influence motivation. The remaining three focused on content and qualities of the application itself and were thought to indirectly influence motivation to use the application.

90x90mm (300 x 300 DPI)

Focus Group Guide – Patient Groups

Introduction

Thanks for taking the time to participate in this study! Today, we're interested in how your Neurofibromatosis (NF1) has affected your pain. We want to hear from you because you are the individuals living with the disease and the pain symptoms.

The information we get from you today will help us customize a mobile application for individuals with NF1. The application has been previously used to help with pain in other diseases. However, before we customize the mobile application, it is important to understand what is needed to make the application as user-friendly and attractive and useful as possible. Thus, we are asking you, the experts, what is needed in this mobile application.

We will be asking you several questions. We would like to set up a few ground rules to ensure we hear from everyone as each of your voices are important to be heard: We ask 1) that you let every individual complete their sentence before you begin speaking, and 2) please say your name before you speak.

As a reminder we will be observing and taking notes. This session will be audio-recorded so that we can go back and review what was said during the session. Please remember you are the experts, and we want to hear your open and honest thoughts.

Do you have any questions before we begin?

---

1. How many of you own a smart phone?
    a. Do you have any experience in using applications to manage any health-related symptoms, i.e. checking your sleep patterns?
    b. If yes, what features do you especially like and dislike in those apps

2. How would you feel about using a smart phone based app to help track and manage your pain related to NF1?
    a. What would motivate you to use it, what key features would it need to have?
    b. What would stop you from using it?

Description of the proposed APP

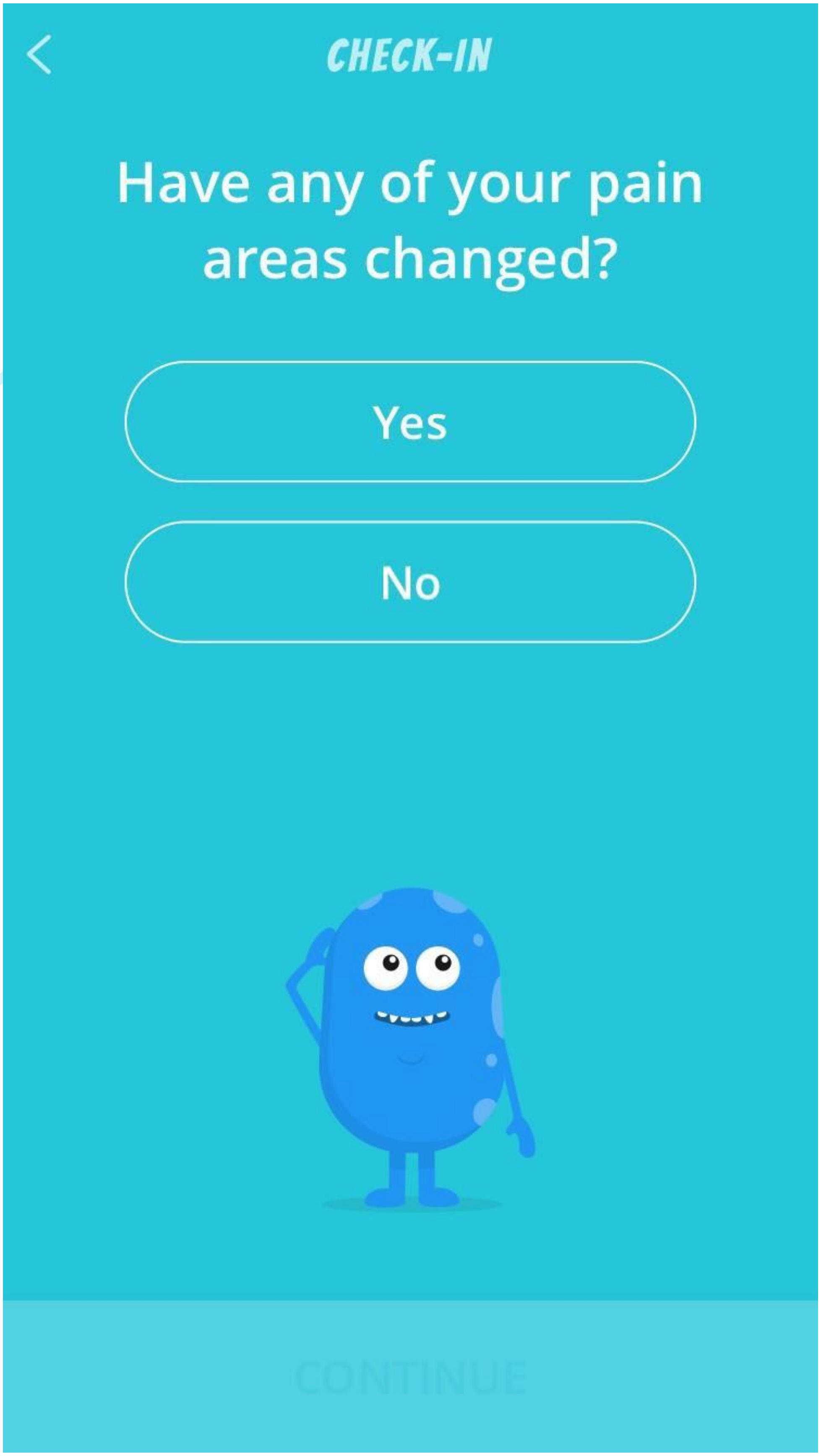
- The mobile application is a customized pain self-management mobile application that is designed to empower people to better manage their pain through symptom tracking, goal setting, coping strategies, and social support. Additionally, the mobile application will provide strategies to reduce pain symptoms through empirically based treatments, i.e. yoga and stretching. **(SEE ATTACHED PICTURES)**

3. What would you envision the mobile application would be like?
    a. What needs to be changed to make it motivating for you to use a pain-related mobile application?
    b. In your opinion, what is the value to have a pain-related mobile application?

4. How likely would you be to use a mobile application specifically for your pain symptoms due to NF1?
    a. Is there a situation where you would use the mobile application more regularly?
    b. What time of day would you be most likely to use the mobile application and why?
    c. If we were going to personalize it for you, what would you have?

5. Is there anything else you want to tell us about your pain experience and using mobile app to help track and manage your pain

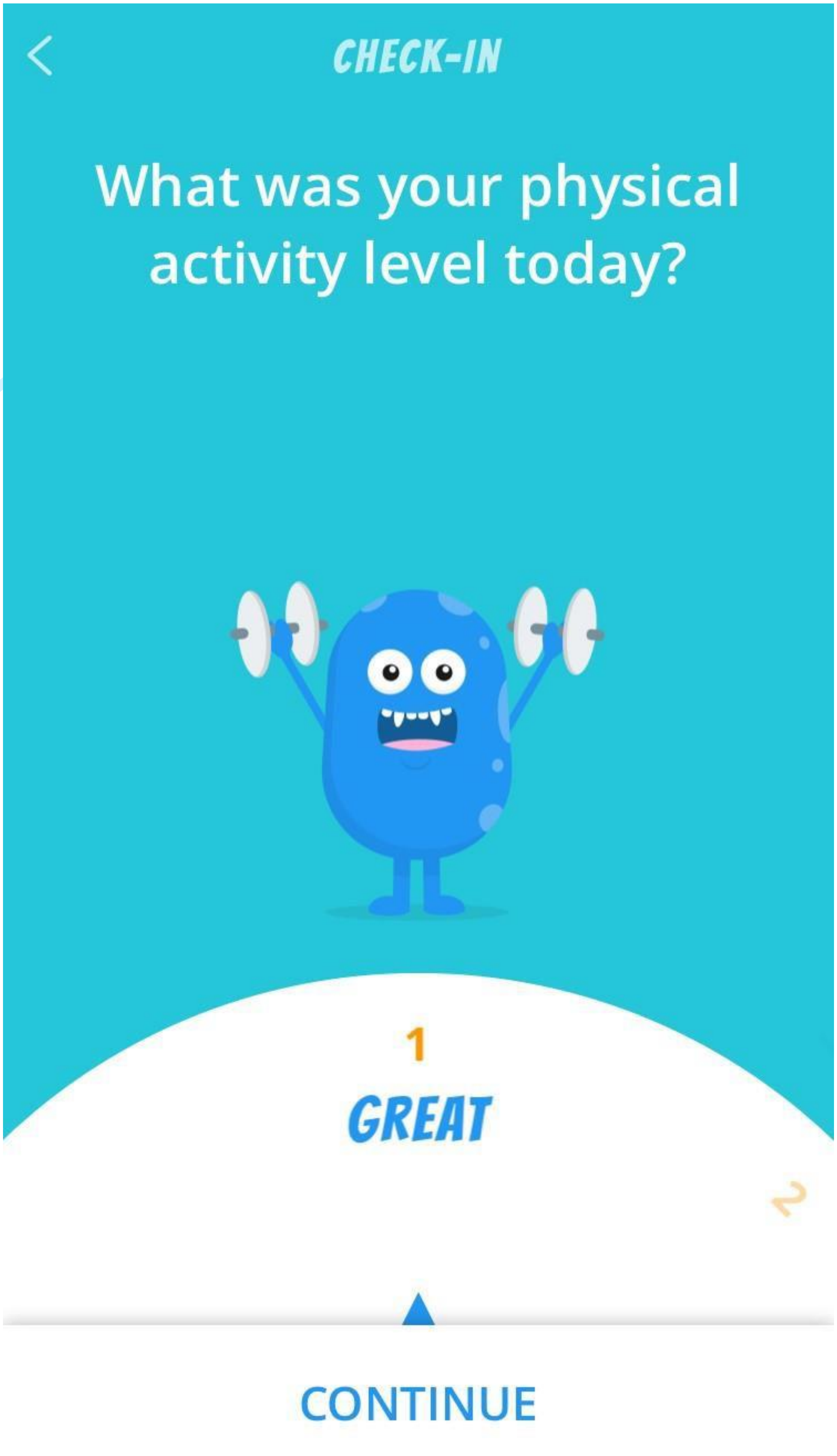
FIRST PICTURE



CHECK-IN

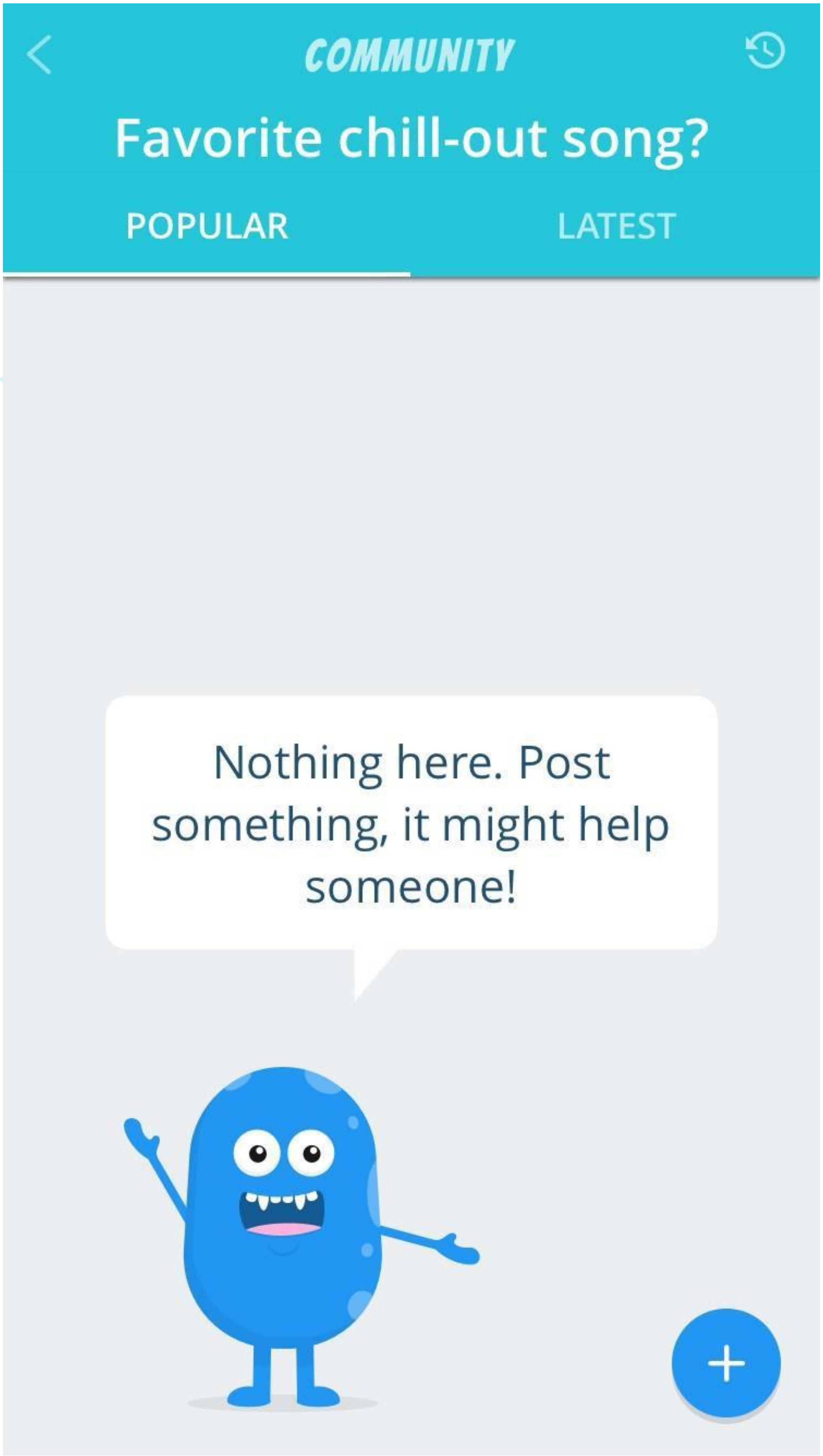
Have any of your pain areas changed?

SECOND PICTURE



CHECK-IN
What was your physical activity level today?
1
GREAT
2
CONTINUE

THIRD PICTURE

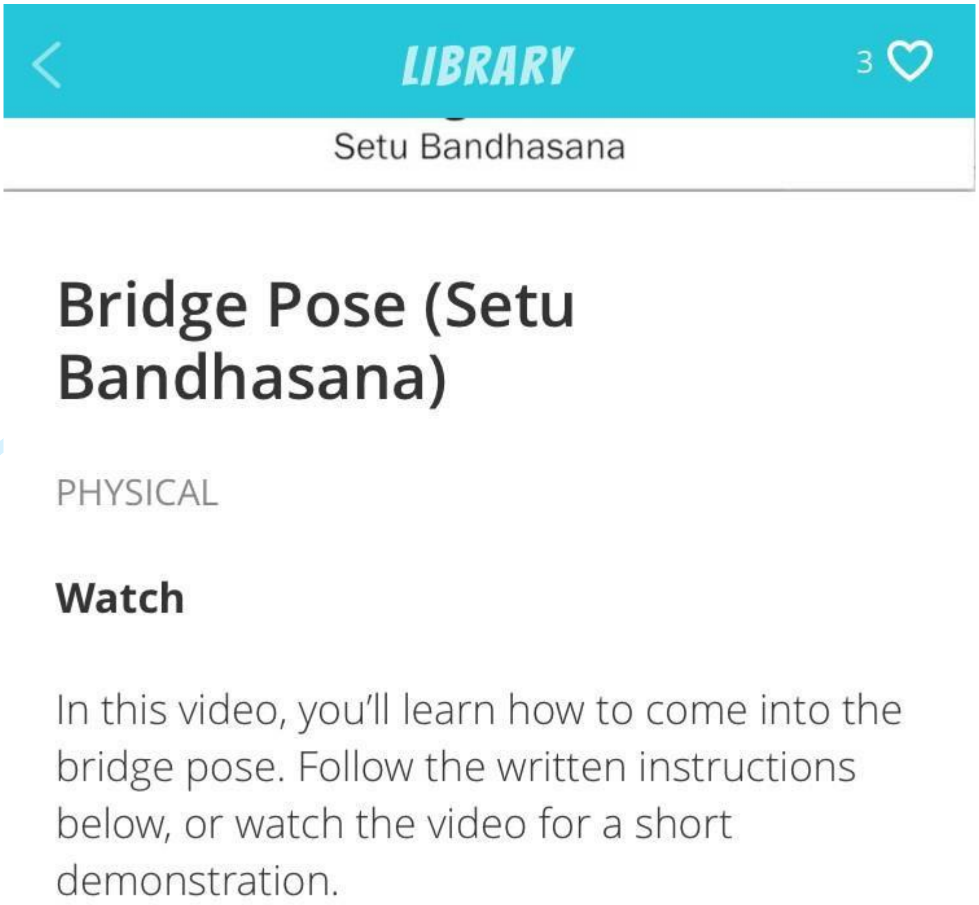


COMMUNITY
POPULAR
LATEST
+

Favorite chill-out song?

Nothing here. Post something, it might help someone!

FOURTH PICTURE



Setu Bandhasana

Bridge Pose (Setu Bandhasana)

PHYSICAL

Watch

In this video, you'll learn how to come into the bridge pose. Follow the written instructions below, or watch the video for a short demonstration.

Focus Group Guide – Expert Groups

Introduction

Thanks for taking the time to participate in this study! The information we get from you today will help us customize a mobile application for individuals with NF1. The application has been previously used to help with pain in other diseases. However, before we customize the mobile application, it is important to understand what is needed to make the application as user-friendly and useful as possible. Thus, we are asking you, the experts, what is needed in this mobile application.

We will be asking you several questions. We would like to set up a few ground rules to ensure we hear from everyone as each of your voices are important to be heard: We ask 1) that you let every individual complete their sentence before you begin speaking, and 2) please say your name before you speak.

As a reminder we will be observing and taking notes. This session will be audio-recorded so that we can go back and review was said during the session. Please remember you are the experts, and we want to hear your open and honest thoughts.

Do you have any questions before we begin?

I want to provide a quick overview of Neurofibromatosis Type 1 (NF1) is an autosomal dominant genetic condition affecting 1 in 2500 individuals. Multiple studies have reported over 50% of individuals with NF1 report significant pain and discomfort which can be associated with tumors, but is often not localized to a structural lesion, thus presenting treatment challenges for patients and their medical caregivers. Due to the complexity of the disorder (e.g., location, severity, number, and type of tumors), there are limited effective therapeutic options for treating pain symptomatology.

1. Can you tell me what strategies do you typically recommend to self-manage chronic pain?
    a. Are they effective in your opinion?

2. Do you have any personal experience in using applications to manage any health-related symptoms (e.g., checking your sleep patterns)?
    a. If yes, what features do you especially like or dislike in those apps?

3. Have you recommended using applications to manage any health-related symptoms (e.g., checking your sleep patterns)?
    a. Did your patients take your recommendations? If no, why not?

Description of the proposed APP

The mobile application is a customized pain self-management mobile application that is designed to empower people to better manage their pain through symptom tracking, goal setting, coping strategies, and social support. Additionally, the mobile application will provide strategies to reduce pain symptoms through evidence-based treatments (e.g., yoga and stretching)**. (SEE ATTACHED PICTURES)**

4. How would you feel about giving a smartphone-based app to help track and manage your clients' chronic pain?
    a. In your opinion, is there value to have a pain-related mobile application?

5. What key features would the mobile application need to have to treat individuals with chronic pain?
    a. What content would be needed to make this helpful for your patients?
        i. Sleep, mood, treatment?

BMJ Publishing Group Limited (BMJ) disclaims all liability and responsibility arising from any reliance placed on this supplemental material which has been supplied by the author(s)

SRQR Reporting checklist for qualitative study.

Instructions to authors

Complete this checklist by entering the page numbers from your manuscript where readers will find each of the items listed below. SRQR reporting guidelines: O'Brien BC, Harris IB, Beckman TJ, Reed DA, Cook DA. Standards for reporting qualitative research: a synthesis of recommendations. Acad Med. 2014;89(9):1245-1251.

		Reporting Item	Page Number
Title			
	#1	Concise description of the nature and topic of the study identifying the study as qualitative or indicating the approach (e.g. ethnography, grounded theory) or data collection methods (e.g. interview, focus group) is recommended	4
Abstract			
	#2	Summary of the key elements of the study using the abstract format of the intended publication; typically includes background, purpose, methods, results and conclusions	5
Introduction			
Problem formulation	#3	Description and signifcance of the problem / phenomenon studied: review of relevant theory and empirical work; problem statement	6-7
Purpose or research question	#4	Purpose of the study and specific objectives or questions	7
Methods			
Qualitative approach and research paradigm	#5	Qualitative approach (e.g. ethnography, grounded theory, case study, phenomenolgy, narrative research) and guiding theory if appropriate; identifying the research paradigm (e.g. postpositivist, constructivist / interpretivist) is also recommended; rationale. The rationale should briefly discuss the justification for choosing that theory, approach, method or technique rather than other options available; the assumptions and limitations implicit in those choices and how those choices influence study conclusions and transferability. As appropriate the rationale for several items might be discussed together.	7-11
Researcher characteristics and reflexivity	#6	Researchers' characteristics that may influence the research, including personal attributes, qualifications / experience, relationship with participants, assumptions and / or presuppositions; potential or actual interaction between researchers' characteristics and the research questions, approach, methods, results and / or transferability	10 & 11
Context	#7	Setting / site and salient contextual factors; rationale	9-10
Sampling strategy	#8	How and why research participants, documents, or events were selected; criteria for deciding when no further sampling was necessary (e.g. sampling saturation); rationale	
Ethical issues pertaining to human subjects	#9	Documentation of approval by an appropriate ethics review board and participant consent, or explanation for lack thereof; other confidentiality and data security issues	11
Data collection methods	#10	Types of data collected; details of data collection procedures including (as appropriate) start and stop dates of data collection and analysis, iterative process, triangulation of sources / methods, and modification of procedures in response to evolving study findings; rationale	10
Data collection	#11	Description of instruments (e.g. interview guides,	10-11

BMJ Publishing Group Limited (BMJ) disclaims all liability and responsibility arising from any reliance placed on this supplemental material which has been supplied by the author(s)

instruments and technologies		questionnaires) and devices (e.g. audio recorders) used for data collection; if / how the instruments(s) changed over the course of the study	10-11 12
Units of study	#12	Number and relevant characteristics of participants, documents, or events included in the study; level of participation (could be reported in results)	10-11
Data processing	#13	Methods for processing data prior to and during analysis, including transcription, data entry, data management and security, verification of data integrity, data coding, and anonymisation / deidentification of excerpts	11
Data analysis	#14	Process by which inferences, themes, etc. were identified and developed, including the researchers involved in data analysis; usually references a specific paradigm or approach; rationale	10-11 & 47
Techniques to enhance trustworthiness	#15	Techniques to enhance trustworthiness and credibility of data analysis (e.g. member checking, audit trail, triangulation); rationale	
Results/findings			
Syntheses and interpretation	#16	Main findings (e.g. interpretations, inferences, and themes); might include development of a theory or model, or integration with prior research or theory	12-22 & 26
Links to empirical data	#17	Evidence (e.g. quotes, field notes, text excerpts, photographs) to substantiate analytic findings	13-22
Discussion			
Intergration with prior work, implications, transferability and contribution(s) to the field	#18	Short summary of main findings; explanation of how findings and conclusions connect to, support, elaborate on, or challenge conclusions of earlier scholarship; discussion of scope of application / generalizability; identification of unique contributions(s) to scholarship in a discipline or field	26
Limitations	#19	Trustworthiness and limitations of findings	25
Other			
Conflicts of interest	#20	Potential sources of influence of perceived influence on study conduct and conclusions; how these were managed	25
Funding	#21	Sources of funding and other support; role of funders in data collection, interpretation and reporting	26-27

None The SRQR checklist is distributed with permission of Wolters Kluwer © 2014 by the Association of American Medical Colleges. This checklist can be completed online using https://www.goodreports.org/, a tool made by the EQUATOR Network in collaboration with Penelope.ai