Applying an integrated knowledge translation framework approach to develop a tablet-based rhythmic movement training intervention for people with Huntington's disease.

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Abstract

Background: Huntington's Disease (HD) is an incurable neurodegenerative disease that leads to the progressive loss of cognitive and motor functions and mood disturbances, largely due to basal ganglia atrophy. Currently, there are no therapeutic interventions tailored to address cognitive and motor impairment in people with HD. Using an integrated knowledge translation (IKT) framework we developed the HD-DRUM intervention, a tablet-based rhythmic movement training application, with the aim to stimulate basal ganglia reliant cognitive and motor abilities in people with HD.

Objective: The primary aim was to develop the HD-DRUM intervention for people with HD. Here we describe the IKT-based process, involving knowledge user engagement, co-design, and iterative usability testing for design refinement.

Methods: The IKT framework was applied to iteratively refine the design of the HD-DRUM application. This process involved three phases of knowledge user engagement and co-design including an online survey into the use of digital technologies and usability testing for design refinement in people with HD. The developed HD-DRUM intervention was described according to the template for intervention description and replication (TIDieR) checklist.

Results: Barriers and facilitators of using digital technologies were identified. Tablets with touch screens were identified as a feasible and accessible platform to deliver the application. Key elements to ensure that the application design and build met the needs of people with HD were identified. The developed tablet-based HD-DRUM intervention can be used at home and allows the quantification of performance changes, remote monitoring of adherence, matching of training difficulty to users' performance levels using gamification, and future scaling-up for reaching a wide range of interested users.

Conclusions: Applying iterative usability testing within the IKT framework allowed the refinement of the design and build of a novel tablet-based intervention to target cognitive and motor functions in people with HD. Mapping the intervention against the TIDieR framework for describing complex interventions, allowed the detailed description of the HD-DRUM intervention and identification of areas that required refinement prior to finalising the intervention protocol.

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Title: Applying an integrated knowledge translation framework approach to develop a tablet-based rhythmic movement training intervention for people with Huntington's disease.

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Keywords: Huntington's disease, eHealth, intervention, training application, rhythm, drumming, movement, cognition, integrated knowledge translation, gamification, TIDieR

Conflicts of interest: JCG received financial renumeration for the recording of the audio and timing content of the HD-DRUM application.

Author contributions: CMB: conception, methodology, analysis, interpretation and writing of paper manuscript, funding; MB, AER, CD, PP: conception, methodology, review and editing of paper manuscript, JCG: creation and design of musical and timing content of intervention, review and editing of paper manuscript, VI: analysis, review and editing of paper manuscript.

ABSTRACT

Background: Huntington's Disease (HD) is an incurable neurodegenerative disease that leads to the progressive loss of cognitive and motor functions and mood disturbances, largely due to basal ganglia atrophy. Currently, there are no therapeutic interventions tailored to address cognitive and motor impairment in people with HD. Using an integrated knowledge translation (IKT) framework we developed the HD-DRUM intervention, a tablet-based rhythmic movement training application, with the aim to stimulate basal ganglia reliant cognitive and motor abilities in people with HD.

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INTRODUCTION

Background and Rationale

Huntington's disease (HD) is an inherited progressive neurodegenerative disease where cell loss in basal ganglia networks of the brain that manifests as cognitive decline, loss of motor control, and mood disturbances. Striatal atrophy [1] and white matter degeneration [2] are observed many years prior to the onset of movement symptoms. These early brain changes are accompanied with impairments in psychomotor speed and executive functions [3, 4] including problems in decision-making, multi-tasking, and motor sequence learning, all of which may hamper a person's everyday functional abilities such as working capacity [5].

Currently, there are no disease-modifying treatments for HD and only very few studies into HD-specific cognition-oriented interventions have been conducted [6]. However, accumulating evidence suggests that neurological music therapy including rhythmic movement and percussion training may be beneficial in the rehabilitation of acquired brain injuries [7-10] and of neurodegenerative disease, including HD [11] and Parkinson's disease (PD), which is another movement disorder that affects the basal ganglia [12-14]. While evidence from high-quality randomised controlled trials is still scarce, recent reviews and meta-analyses concluded that music interventions may be beneficial for gait, timing of upper extremity functions, and quality of life after stroke [7], may improve gait and mobility in Parkinson's disease [12] and motor and cognitive functions in HD [11].

Previously, in two pilot studies we explored a rhythmic movement training (Bongo drumming) as a therapeutic tool for people with HD [15, 16]. We chose drumming because it is an activity that requires the learning, planning, and execution of movement sequences, which are all abilities that depend on basal ganglia functions and become more difficult as the disease progresses. Thus, we hypothesised that drumming would not only improve motor abilities and response speed, but also cognitive functions involved in the planning and execution of movements and multi-tasking.

In our pilot research [15, 16], people with HD followed audio instructions teaching them to drum along on a pair of Bongo drums. Each 10-15 min training session introduced a novel drumming pattern, starting with simple, slow patterns and gradually increasing in complexity and speed. We found that 2 months of drumming training at home was acceptable for people at premanifest and early stages of the disease and was associated with significant improvements in cognition and white matter microstructure. Anecdotally, some patients and their spouses reported benefits in hand coordination and general alertness. However, the training delivery on Bongo drums also had some disadvantages. Firstly, it did not allow the quantification of training-induced performance improvements nor the recording of adherence, i.e., the duration and frequency of training engagement. Secondly, as drumming responses were not tracked, the level of training difficulty could not be matched to an individuals' performance level to avoid over- and underchallenge. Thirdly, delivering the training on CDs and Bongo drums makes an increased scale of use more challenging compared with the wide accessibility of digital applications on tablets and smartphones.

An electronic health application of the drumming training that can be delivered on smartphones or tablets has the potential to address these issues. However, there is very little known about the use of digital technologies and any potential barriers in people with HD with the exception of research into wearable or portable sensors to monitor motor and cognitive alterations in HD [17, 18]. A recent review of the literature concluded that digital technologies hold promises for therapeutic research and symptom management of HD but that devices need to be standardized and protocols harmonized to optimize their clinical use in HD [17].

Electronic health applications (eHealth) are increasingly employed as cost-effective and widely accessible tools to support health care delivery, monitoring, and education [19, 20]. With the recent explosion of the eHealth market, ensuring that eHealth applications are appropriately designed and meet the needs of the end-users before employing them as health interventions has become increasingly important. Similarly, it has been recognised that published descriptions of eHealth interventions often lack sufficient detail to replicate or evaluate their effects.

Recently, the UK Medical Research Council (MRC) and National Institute for Health Research (NIHR) have updated their framework for the development and evaluation of complex interventions including eHealth [21]. The new framework recommends engagement of stakeholders, addressing uncertainties, refinement of intervention and theory, taking into account the context of intervention delivery, and economic considerations as core elements to guide research into complex interventions [22]. These recommendations reflect a growing support within health research for co-design; that is the engagement of knowledge users in the development and evaluation of interventions [23]. Knowledge users refer to any stakeholders involved in influencing, administering, and/or using the health care system, including those with lived experience. One framework developed to facilitate co-design is the integrated knowledge translation (IKT) approach which refers to an interactive process of knowledge exchange between different stakeholders to produce interventions that are useful to health care system knowledge users [24]. Within the context of eHealth, the US Institute of Medicine has identified usability (the extent to which an end-user can use the product to achieve specified goals) as a key component of good practice for the development of electronic applications [25]. Therefore, the usability of an eHealth application needs to be tested by knowledge users as part of the developmental process to ensure that end-users' needs are met. Thus, engagement of knowledge users in co-design and iterative usability testing for design refinement have emerged as important components of an IKT approach for the development of eHealth interventions [26].

We adopted the IKT framework for developing a tablet-based rhythmic movement training application intervention for people with Huntington's disease. This paper first reports the IKT-based intervention development process and then provides a detailed description of the intervention using the TIDieR checklist [27].

Objectives

The primary aim of this research was to develop HD-DRUM, a tablet-based rhythmic movement training application intervention for people with HD.

METHODS

We developed a proof-of-concept tablet-based drumming training app, HD-DRUM, ready for clinical evaluation. The purpose of the HD-DRUM intervention is to provide a digital training platform that provides solutions to the challenges encountered with the use of the Bongo drumming training that we investigated previously. Specifically, it allows (i) quantification of performance improvements, (ii) quantification of training engagement, (iii) matching of the training difficulty to performance levels, and (iv) scaling-up reach for a wider audience via delivery on tablets and/or smartphones.

Following the IKT framework, and in collaboration with the user-centred design and innovation agency Kinneir Dufort (KD)[28] and knowledge users, we applied a dynamic and iterative model of development that included knowledge-user engagement at different stages of the planning and development of HD-DRUM (Figure 1).

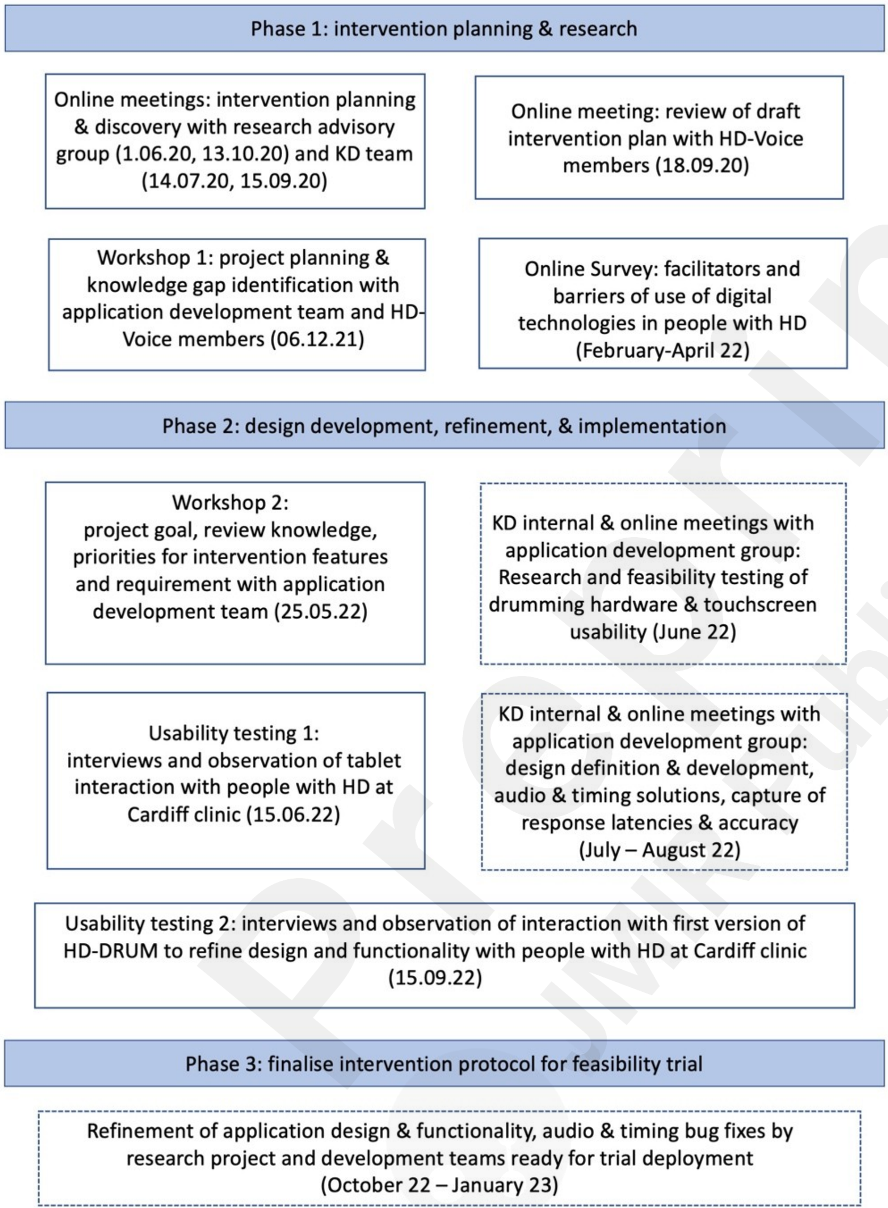

Figure 1. Overview of the integrated knowledge translation approach of the intervention development process. Abbreviations: HD = Huntington’s Disease, KD = Kinneir Dufort.

Participants and Recruitment

For this study the following four groups were formed:

Application (app) development group
comprised of the lead researcher (CMB), the KD design and engineering team, and musician JC.

This was the core team that designed and build the HD-DRUM application in collaboration with the knowledge users and the research advisory group.

Research advisory and management group
comprised of an interdisciplinary team of researchers, experts, and clinicians working in the fields related to the project and research area. This group consisted of five researchers (CMB, AR, CD, MB, PP) with expertise in psychology, neuroscience, intervention development and evaluation, clinical trial methodology and statistics, and neurology. This group was established prior to starting the IKT development process to help plan the intervention development, approach, research questions and outcome measures, and to monitor the progress of the project in line with milestones, on a monthly basis.

Knowledge user group
comprised of people affected by HD, including people with HD, their carers, and family members and clinical staff working in the HD field. Five people affected by HD were members of HD-Voice, the patient and public involvement group of the UK Huntington's Disease Association (HDA), a third sector support organisation. Twenty-nine individuals with HD were recruited *via* an advert in the HDA newsletter and participated in an online survey about facilitators and barriers of the use of digital technologies. Demographic information of age, gender, education, and disease stage for these participants was collected as part of the online survey. In addition, knowledge users were recruited for usability assessments from the Cardiff University HD clinic. Seven people participated in a usability assessment of tablet interactions in people with HD. These were four people with early to moderate manifest HD, two carers, and one clinician. Another four people with HD at various stages of disease progression (premanifest to manifest) and one member of the clinical staff took part in usability testing of the first version of the HD-DRUM app.

Research project team
comprised of three core research members responsible for running the feasibility trial into *HD-DRUM*. This group consists of the lead researcher (CMB), the postdoctoral research associate (VI) and a PhD student and was established during the finalisation phase of the app development. Members of this group piloted and tested the app prior to trial deployment.

Intervention development

Phase 1: Intervention planning, research, and feasibility

Members of the research advisory group met virtually online and communicated via email to review an updated project plan based on the findings of the previous pilot studies[15, 16]. They discussed the research questions, the research approach, and outcome measures of the updated project in preparation for a funding application. In parallel, CMB engaged with the KD team to develop a road map for the design of the new intervention app that incorporated knowledge user engagement and iterative usability testing throughout the development process. The project plan was reviewed in an online meeting between the lead researcher and members of HD-Voice, who provided feedback on the feasibility of the planned intervention and research protocol in people with HD. To minimise participant burden and increase retention rates, it was suggested to shorten the originally proposed training duration of 6 months to 2 months and to limit assessments to before and after the intervention and at corresponding time points for the control group in the feasibility trial
. It was also suggested to develop a stand-alone application version for offline use to include those individuals without an internet connection in the research. This feedback was incorporated into the research protocol.

Once funding was secured for the project, the app development team met in a hybrid workshop with members of HD-Voice to review the project plan and to identify uncertainties and knowledge gaps that needed clarification prior to designing an app solution. Specifically, the need to gain an understanding of how people with HD use digital technologies, what barriers they encounter, and what design and functionality requirements needed to be met when designing and building the app was identified. Given the lack of evidence in the literature, we conducted an online survey into facilitators and barriers of use of digital technologies in people with HD.

Online survey into the use of digital technologies in people with HD

The survey consisted of 21 items and was delivered and analysed on Qualtrics (version 2022) [29]. Items included (i) the frequency of use of different digital devices (mobile phones/smartphones, tablet, computer/laptop) and the internet, (ii) the preferred digital device, (iii) the type of digital activities carried out (emails, social networking, buying goods/services, listening to music, downloading information, games, images, video calls), and (iv) how confident participants felt in conducting these activities as rated on a 5-points scale ranging from “not at all” to “very confident”. Further, participants were asked to provide information as to whether they required any assistive technologies or accessibility settings and whether they had encountered any problems when using digital devices.

Phase 2: Design development, refinement, and implementation

At the start of this phase, the development team and a member of the research advisory group met to align all team members on the project goal, review and build knowledge including a discussion of the results of the on-line survey. In addition, prioritisation of various app components was discussed. The KD team then researched different options for hardware and software configurations suitable for a drumming application including drumming on a tablet touchscreen and drumming on connected or standard bongos. Direct drumming on touchscreens had the advantage of reducing costs, complexity, and technical risks (from additional circuitry and choice of audio transducers) but required usability testing with people with HD.

To assess the feasibility of tablet interactions in people with HD, we engaged with service users of the Cardiff based HD clinic. The freely available bongo drumming application *Congas & Bongos*[30] was installed on two different tablets of varying screen sizes: a 10inch Samsung Galaxy and a 12.9inch iPad Pro. For comparability purposes the “four and five finger swipe multi-tasking gesture” on the iPad Pro was deactivated prior to the test. Participants were approached in the clinic waiting room and encouraged to interact with the bongo drumming application on both tablets. The tablets were placed on a table in front of the participants. Participants were asked which screen size they preferred. The research team made notes of any feedback and suggestions from participants and any observations on how people interacted with the application.

The application development group then started the design process, which involved close collaboration between the KD team, JC who recorded the audio and timing content, and CMB who monitored alignment with project goals and specified requirements. Core elements of the development process concerned the application design, the implementation of its audio and timing features as well as response latency and accuracy capture. During the design definition and development phase, different design solutions were iteratively tested and refined internally by KD to ensure accessibility, functionality, usability, and delight. Different solutions for audio and timing specifications and latency capture were investigated and evaluated with regards to functionality, technical risk, and cost effectiveness.

The first version of the HD-DRUM application was then tested for its usability with people with HD in the Cardiff HD clinic. Usability testing took place in the waiting area of the Cardiff HD clinic to refine the application design to the needs of people with HD by observing how participants interacted with the application and by gaining feedback and suggestions about what worked well and what needed to be improved. The tablet was placed in front of participants on a table sufficiently close to them so that they could reach the tablet comfortably and participants were encouraged to interact with the application. Researchers observed these interactions involving opening and navigating through the application and engaging with the first training sessions, made notes, and collated feedback.

Phase 3: Finalisation of the intervention protocol for feasibility trial

Design and functionality refinements based on the usability findings were implemented. The application was iteratively tested and corrected for any remaining bugs including audio and timing errors by the project research and development teams. The end result was the HD-DRUM application intervention outlined and described in Table 2.

RESULTS

Digital technologies survey

Participant demographics

Fifteen female and 14 male individuals over the age of 25 years (67% between 35 and 65 years) completed the survey anonymously (Figure 2). Half of those individuals were university-educated at undergraduate (21%) or postgraduate (31%) level including 7% with a doctoral degree. Of the remaining participants, 21% had A-level/BTECH, 14% GSCE, and 14% other educational/vocational qualifications. Most participants reported being at the premanifest (28%) or early disease stages (58%) while 14% reported middle or later stages.

Use of digital technologies, facilitators, and barriers

Figure 2 displays the main results of the online survey. Most participants (86%) reported using the internet on a daily basis (83% multiple times), and 7% used it once a week or once a month (Figure 2A).

The most popular devices were smartphones (55%) and tablets (31%) (Figure 2B) with most participants using them every day (86% for smartphones, 34% for tablets). PCs and laptops were used by 51% at least once a day, but only 10% preferred them over other devices.

Participants who used digital devices and the internet, engaged in a range of activities including sending/receiving emails, social networking, purchasing goods and service, reading the news, downloading games, images, films, news articles, playing music and participating in video calls (Figure 2B). Overall, the majority felt fairly or very confident in carrying out these activities ranging from 62% for downloading news and social networking to 81% for listening to online music (Figure 2C).

However, some participants reported that they never used a device (52% for tablet, 21% for PC/laptop, 10% for smartphone) or the internet (7%) and did not feel confident in carrying out the above activities (ranging from 4% for listening to online music to 10% for participating in video calls and social networking).

Similarly, while most individuals reported that they did not require any assistive technology or accessibility settings (82%), two participants mentioned that they used password reminders or did not know what assistive technology was available (Table 1). Table 1 lists the problems participants experienced when using digital technologies. These difficulties seem to become apparent with the development of clinical symptoms as all participants who reported barriers were at manifest disease stages, but older age may also have contributed to them.

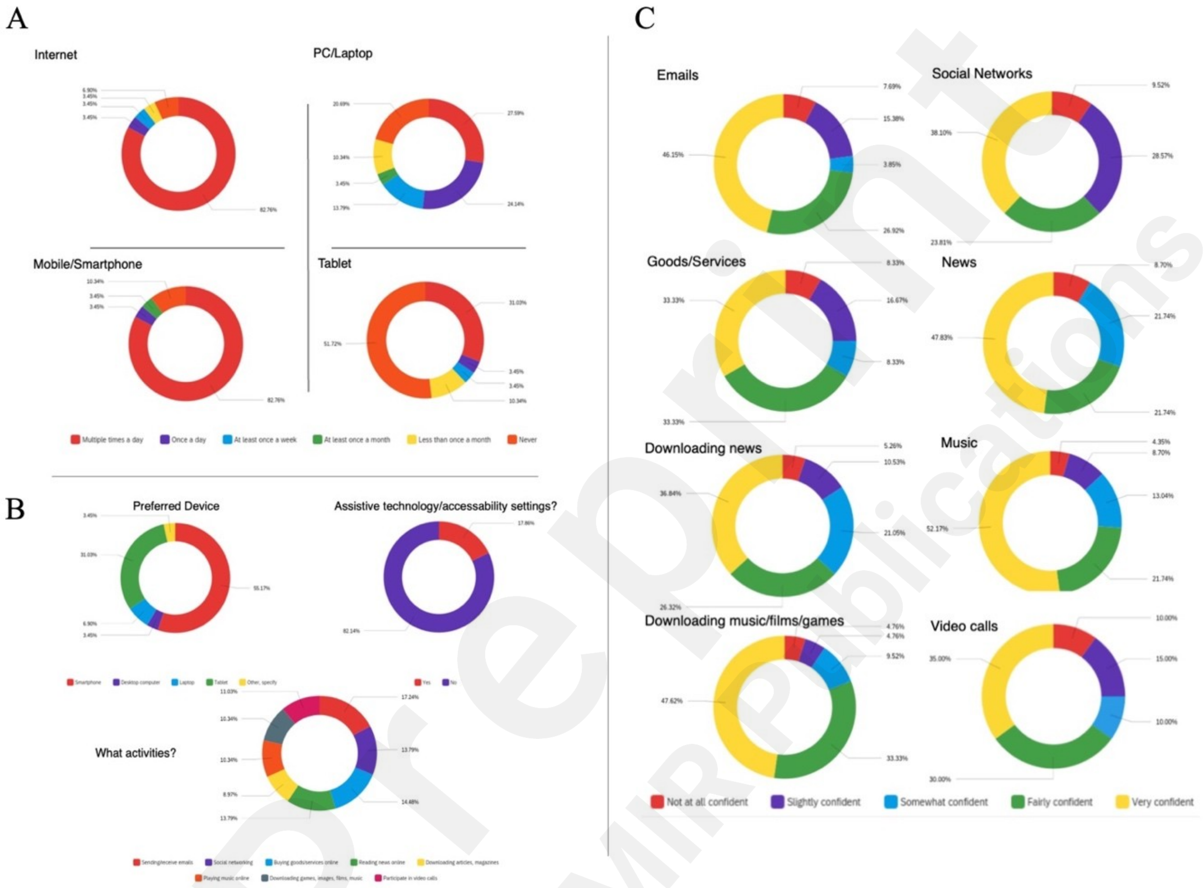

Figure 2. displays the main findings of the online digital technology survey from n = 29 individuals with Huntington's disease. A) Use of different devices, B) preferred device, what activities used for and requirement for accessibility settings, C) Confidence ratings for individual activities.

Table 1. Barriers to using digital technologies reported by survey participants

HD stage	**Assistive technology/ accessibility settings**	**Problems**
early		
	Password reminder	I can find it difficult to understand what is being asked at times or can't find information. I can take a long time to find applications that I use on my iPhone

		frequently.
	-	I struggle with a mouse on the computer so bought a tablet instead
	-	Yes, accidentally clicking on the incorrect letters and words when using automatic texting. I don’t use voice activated smart devices like Siri as already fed up repeating myself with people
	-	I have to enlarge the page and it takes me so long to type, it’s very frustrating
	-	I have to type really slow, often people don't think I’m going to answer
	-	Problems with online banking - my wife helps me
	-	Find it hard typing on small keyboard
later		
	I don’t know what is available.	My lack of dexterity and grip mean that I am unable to operate any electronic/digital devices.
	-	I can read but not send emails. I cannot use applications.

Summary and conclusion of survey results

In summary, the responses to our survey suggested that most participants were engaging with digital technologies and using smartphones, tablets, and PC/laptops for numerous online activities. However, the responses to our questions about problems with using applications identified some accessibility issues that would need to be addressed in the application design for people with manifest HD.

Usability testing of tablet interactions in people with HD

We tested our assumption that interactions with tablet touch screens of two different sizes were feasible for people with HD with people with HD at the Cardiff clinic.

Screen size and position

We found that both the large and small tablets performed well in terms of participants being able to tap on the virtual bongo drums. Participants did not indicate a preference for one or the other screen size. However, when asked some participants preferred the larger device due to a difference in screen and sound quality. Multiple participants said they own a tablet but not all were engaging with it. One participant who took part in the original Bongo drumming pilot study said that they preferred the tablet to the physical bongos. More important than screen size was the placing of the tablets close enough for people to reach. Multiple participants moved the tablets closer to them because reaching too far out led to more mis-presses.

Interaction with the application

Participants used their index finger or index, middle, and ring fingers together to tap on the tablet. One participant with manifest HD alternated between one and multiple fingers due to limited ability to control the movement. For some people with manifest HD initiating the first tap appeared to require a lot of effort and concentration. However, once started the following interactions appeared easier. Some impulsive and perseverative responses were observed, i.e., the same tapping interaction was repeated with minimal control of speed or force. The audio feedback upon tapping on the virtual bongos was positively received. The application used showed drums in two different sizes, but it was agreed that this was not particularly necessary for our project. Multiple participants appeared joyful whilst playing and their body language demonstrated comfort when immersed in the drumming interaction.

Motivation and engagement

Multiple participants mentioned the need to gamify the application to maintain engagement. Some participants played computer games and stated that the application would have to be more engaging than games such as Solitaire. Multiple participants also mentioned the need to have music alongside the drumming to keep the training interesting and engaging. The use of headphones that could be plugged into the application was received well as it helped with more focus and would prevent family members from listening in or being bothered by the noise. A family member mentioned the potential mental health benefits of engaging in the drum playing. Multiple participants thought it would be helpful to be reminded to drum on the application as a method of engagement. Fifteen-minute sessions were generally accepted as a feasible length, but participants were hesitant to suggest this was a comfortable length of time. A participant discussed with their carer(s) that it would be fun to be able to play together, perhaps alongside other instruments.

Outcome of tablet usability testing

Successful drumming on the screen of both tablet sizes suggested that a tablet-based version of the application using virtual bongos would be feasible for use by people with HD. The smaller Samsung Galaxy 10inch was chosen as a more cost-effective and therefore accessible hardware solution than the iPad Pro. It was evident that provision of clear instructions regarding the tablet position on the table and the hand gesture used for tapping on the virtual drums (playing with the tips of index, middle, and ring fingers) to facilitate interaction with the application was important. User testing indicated the addition of reinforced screen protection and bumper cases for improved durability was necessary to mitigate difficulties with controlling the force and speed of tapping. Further, an element of gamification and positive feedback should be included in the application design as well as the playing to background music to keep the training engaging.

Usability testing of the HD-DRUM application

Usability testing of the first version of the HD-DRUM application was undertaken to gain feedback for refining the interface to ensure accessibility, functionality, usability, and delight.

Interaction with the application

All participants were able to use the application and navigate through it. One participant with manifest HD required one-to-one instruction initially but then appeared to progress afterwards. Participants enjoyed the voice over by musician (JCG) and the look and feel of the interface which was called “clear’. Overall, participants appeared happy and joyful while interacting with the application.

All participants understood the task of drumming along to the audio instructions. Participants also seemed to understand the different expectations between training and performance parts of the intervention and were able to complete the performance sessions. A visual halo cue alongside auditory feedback seemed to help further identify the beat on which to drum; specifically expressed by some participants and simply observed in others. The addition of background music provided a good challenge for some participants, one of them noting that it made them concentrate more. The training session length of about 10-15 min was considered acceptable. Longer training sessions would not be acceptable.

Participants commented that they enjoyed the element of gamification by having to reach a certain success level to unlock the next training session and the positive feedback at the end of the session that congratulated them for having completed the session.

Refinements to the application content and instructions

Participants reported that the introduction session explaining how to use the application was too long and detailed with some confusion about whether they were supposed to drum along during the introduction or not. Therefore, the introduction session was shortened and made more focused and concise.

Some people with manifest HD found the first training rhythm of 65 beats per minute too fast as an introduction. They tended to tap on all 4 beats instead of the single beat as instructed. They were also confused between the spoken metronome and beat. In response, we included a slower session of less than 1 beat per second with regular tapping on all 4 beats without the metronome as the introductory session.

Participants predominantly used single fingers over flat hands and needed explicit instructions to tap on the virtual drums with a flat hand using all fingers together to mimic Bongo drumming. To compensate for this, the instruction manual for the application includes detailed instructions on how to tap correctly. Participants will also receive face-to-face instructions and demonstration by the research team at the start as part of the intervention delivery.

Refinements to the application design

Home/session screen: When participants interacted with the session icons on the screen (Figure 3B), they expected that tapping on a full size, centred icon would start the session and that tapping on a minimized icon would scroll and centre that session instead of using the arrow and play buttons at the bottom of the screen. These functionalities were therefore added to the application design. The introduction session was not recognised as different content to the training session and was therefore made visually distinctive with an intro label. The session icon artwork was grouped in threes with regards to colour and shape to reflect the background track being the same for three sessions in a row.

Pause screen: The pause screen caused confusion and there was a risk of participants accidentally pressing “exit” or “restart” instead of “resume” and lose their progress as a result. Consequently, the design was changed to a more prominent (in size and location) resume button to reflect the fact that resume was the main action, and restart and exit were secondary action choices. Further, a confirmation step after pressing “restart” or “exit” was added to avoid users making these choices by mistake.

Drum and end of session screens: Static icons were replaced with dynamic ones to reflect the session that was playing and to allow the user to finish the session by either tapping the icon of the session just played, if this was to be repeated, or by tapping the icon of the newly unlocked session.

Summary

Overall participants were positive about the application design and the training content. They were able to navigate through the application and follow the audio instructions. They appreciated the visual halo cues and auditory feedback when tapping the virtual drum as well as playing along to background music and the gamification element of the application. However, some refinements to the design and functionality of the screen displays, the length and content of the introduction and the first training session, in addition to more detailed instructions with regards to the tapping responses were required to maximise accessibility, functionality, usability, and delight of the interface.

Table 2. Description of the HD-DRUM application (version 1.0) intervention according to the TIDieR framework

Template for Intervention Description and Replication (TIDieR) item no	**Description**
1. Name/Description of intervention	
	HD-DRUM, a tablet-based drumming training application intervention aimed at stimulating cognitive and motor abilities in people with Huntington's disease (HD).
2. Why: Rationale, theory, goal(s)	
	Rational: Huntington's disease (HD) causes neurodegeneration in the basal ganglia leading to a progressive loss of cognitive and motor control. Drumming involves the learning of rhythmic motor sequences and our pilot research suggested benefits of Bongo drumming on cognition and neural pathways in people with HD [15, 16]. The holistic evaluation of the

	potential therapeutic benefits of drumming in HD requires a digital training solution that allows (i) the quantification of performance improvements and training engagement, (ii) the matching of the training difficulty to performance levels to avoid over- or underchallenge and (iii) scaling-up to reach a wider audience via delivery on tablets and/or smartphones. The HD-DRUM application has been designed to address these points and to meet the accessibility needs of people with HD.
3. & 4. What: Materials & Procedures	
	The HD-DRUM application has been programmed in native Android Java and runs on the Android operating system (version 21 or higher). The chosen hardware is a Samsung Galaxy Tab A8 with a 10.5inch screen. Participants are provided with the application on a tablet in a protective case and with a 17-page instruction manual. Participants start the application by tapping on the HD-DRUM icon on the tablet home screen. HD-DRUM consists of 23 audio recordings, one introduction and 22 training sessions each 10-15 min long. Each training session introduces rhythmic patterns that are based on paradiddles and different rhythmic styles including Hip-Hop, Funk, Samba, and Reggaeton. Patterns are practised with and without a metronome and/or background music. The shape and colour of each session's identity is unique and reflect the session's rhythm and background track (Figure 3B). The first three sessions introduce slow and regular rhythms and are available to all users right from the beginning. The patterns then gradually increase in complexity and tempo through the program. After starting the application, the introduction and the first three sessions and any unlocked sessions are displayed on the HD-DRUM home screen and participants can scroll through them (Figure 3B). A session is active and can be started when its circle icon is displayed enlarged in the centre of the screen. A session can be started by either tapping on the session icon or by pressing the start button. When a drum session is opened, two virtual drums, a blue triangle and red circle appear in transparent colours on the screen and need to be tapped to start the audio instructions (Figure 3C). Alternatively, the start button can be pressed. Participants can pause the session at any time. When the session has been paused there are three options: resume, restart, or exit the session (Figure 3D). A bar on the top of the screen shows the participants how far they have progressed through the session. In each training session, participants are instructed to tap

	or drum along to the audio instructions on the two virtual drums (the blue triangle and the red circle) on the tablet screen. They are told to tap with the tips of all fingers simultaneously. The virtual drums mimic physical Bongo drums by producing visual (shrinking) and audio feedback (a high pitch Bongo sound for the left triangle and a low pitch sound for the right circle) when tapped. In sessions 1-7 the rhythmic patterns are first practised with each hand separately and then with both hands together; after session 8 all patterns involve both hands and are practised starting with one hand and then in reverse order with the other. The application includes an element of gamification such that from session 3 onwards, participants must reach an accuracy level of 70% or higher to unlock the next more difficult session. If their performance accuracy falls below this threshold, they are encouraged to repeat the session or practise one of the previous sessions. At the end of each session, participants receive positive feedback for either having completed the session (for accuracy levels below 70%), or for having unlocked the next session (for accuracy levels larger or equal 70%) (Figure 3E). Each session consists of training and performance parts. During the training, participants learn and practice to tap along to a new rhythmic pattern. During performance parts, participants are asked to tap or drum along as accurately, i.e., as synchronised as possible with the recorded Bongo sounds, while their response accuracy and latencies are recorded. To facilitate performance, drumming is guided by a visual halo cue that appears around the target triangle or circle when a hit is expected (Figure 3C). The interaction with each training session produces an output file in comma-separated format that is stored on the tablet and uploaded to a project-specific space on Google Firebase (https://firebase.google.com) when the tablet is connected to the internet. The output files contain a user ID code and time stamp information about session engagement, expected hits and timing and accuracy of responses. Summary statistics are provided for the total number of drum hits in the session track, the allowed time window in milliseconds for correct responses (see below), the success accuracy threshold, information about the date, time, and duration of engagement, average hit accuracy, left and right-hand hit accuracies, average reaction times in milliseconds, and average and best reaction times for left and right-hand responses.

	Each session audio file is accompanied by a Musical Instrument Digital Interface (MIDI) file that contains information about the timing of the audio drumming sounds. Accuracy levels are determined by comparing the time stamps of the sounds in the MIDI file with the recordings of participants' responses to the audio instructions. An accurate response in HD-DRUM is defined as a correct left or right-hand tap within a predefined, symmetrical time window around the expected hit stored in the MIDI file. Based on previous findings of simple and choice reaction times in HD [31] and our own usability testing in the Cardiff clinic, the largest time window for the slowest rhythmic pattern (less than 1 beat per second) in session 1 was set to 1,000 milliseconds, i.e., 500 milliseconds before and after the expected hit. Time windows were then gradually reduced with increasing tempo to 100 milliseconds (50 msec before and after expected hit) for rhythmic patterns with tempos over 100 beats per minutes (maximum 117 beats per minute).
5: Who provided	
	The Cardiff University research team provides participants with the application for home training.
6: How delivered	
	Participants engage with the training on an individual basis at home. If possible, family members and/or carers will be engaged to facilitate the training by providing a supportive environment. The research team monitors adherence and progression remotely and stays in weekly contact with participants via email, text, and/or phone calls.
7: Where delivered/required infrastructure	
	The stand-alone tablet-based training is delivered in participants' home. Participants are instructed to place the tablet flat on a table in front of them at a comfortable reaching distance. Internet connection is not required for training delivery.
8: When & how much	
	Participants are instructed to practice for ~10-15 min per day, 5 times per week, for 8 weeks.
9: Tailoring	
	From session 3 onwards, participants must reach an accuracy level of 70% or higher to unlock the next more difficult session. If their performance accuracy falls below this threshold, they are encouraged to repeat the session or practise one of the previous easier sessions.
10: Modifications	
	The current version of HD-DRUM is a proof-of-concept

	prototype. The feasibility evaluation will inform future modifications to the application.
11: How well: Planned	
	Adherence with the training is remotely monitored by the research team via the output files that are generated during application engagement and uploaded onto Google Firebase when the tablet is connected to the internet. The research team also keeps in regular contact with participants.
12: How well: Actual	
	The feasibility assessments are on-going.

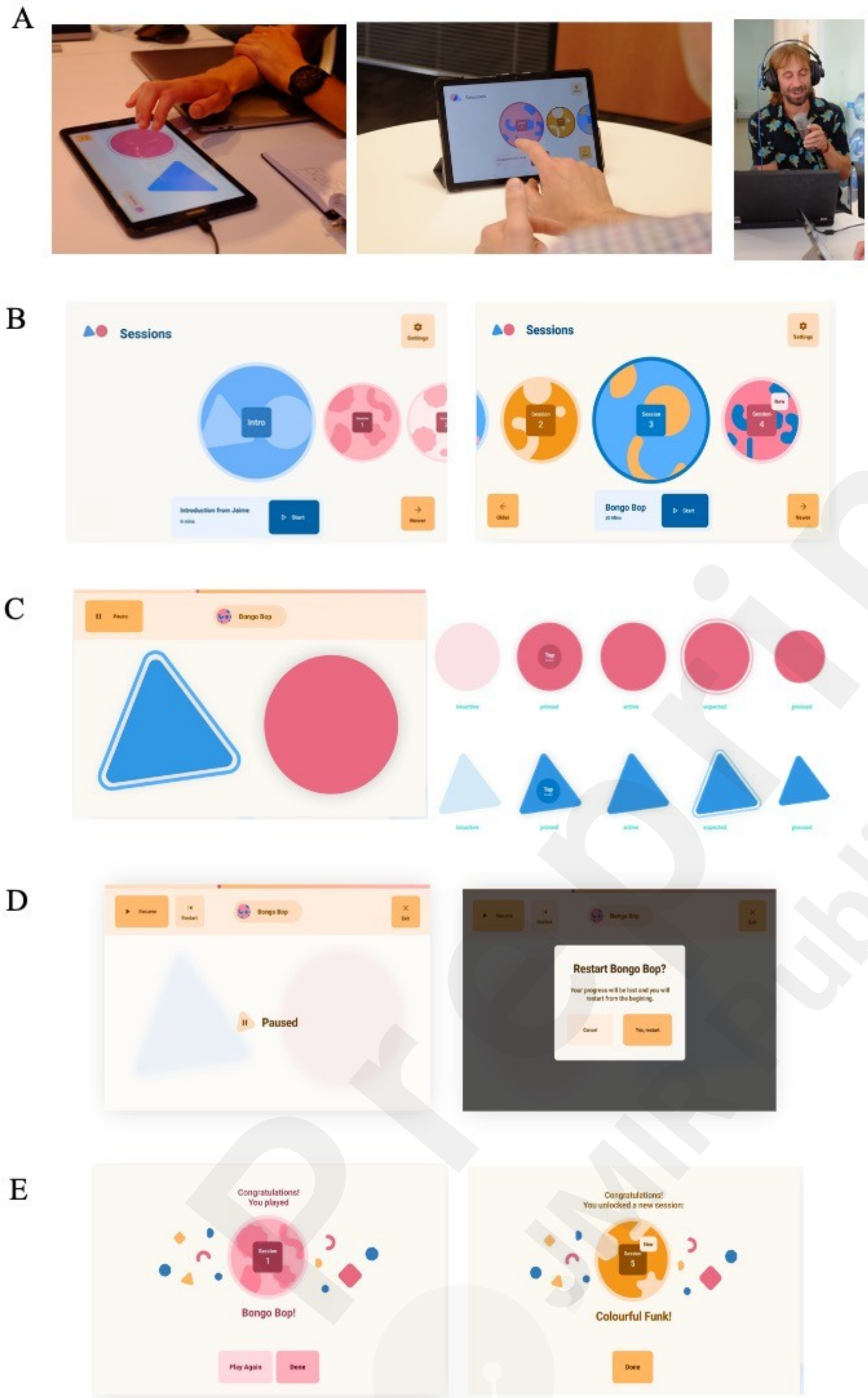

Figure 3. gives an overview of the HD-DRUM application design. A) The HD-DRUM application comprises 22 audio training session that were recorded by musician JC and runs on a Samsung Galaxy 8 tablet. B) The home screen displays the introduction and all unlocked training sessions. Users can scroll through and start sessions. C) The drum screen displays two virtual drums, a blue triangle and a red circle that respond with high and low pitch Bongo sounds when tapped. A visual halo cues the expected hit during performance sessions. D) Training can be paused at any time. A confirmation screen appears when participants select restart or exit the session prior to completing it. E) At the end of each session user receive positive feedback congratulating them on having completed the session or for having unlocked the next session.

DISCUSSION

Here we have described the codesign and refinement of a tablet-based drumming intervention application for people with HD. We used an IKT approach for the intervention development process that involved knowledge user engagement and iterative usability testing. The three phases allowed identification of knowledge gaps and changes and adaptations required to address accessibility needs of people with HD. We have described the resultant intervention according to TIDieR to provide details of the intervention's rational, key elements, design, and functionality to enable future replication.

The IKT process enabled an inter-disciplinary research team to engage with a design company, musician, and knowledge users in a semi-formalised way to formulate, co-design, and refine an intervention application suitable for clinical evaluation. More specifically, the application addresses the methodological shortcomings of our previous pilot research into the feasibility of a Bongo drumming intervention in people with HD through the quantification of training progression and adherence, the matching of performance to training difficulty, and the potential for increased scale of use. This allows the objective assessment of the feasibility and the effects of HD-DRUM on rhythmic movement performance and the scaling up of sample sizes in future randomised controlled trials. Matching users practise to a level appropriate to their abilities is expected to increase acceptability of the intervention by avoiding frustration and boredom due to over- and underchallenge.

By involving people with HD in the application development process, we were able to identify and address accessibility barriers and refine the application design to maximise accessibility, functionality, usability, and enjoyment of the interactions with the interface. Codesign highlighted the importance of designing the application in a user-friendly and intuitive way with a focus on key functionalities of the application. This was achieved by allowing navigation through the application with large and visually distinctive buttons on a tablet-based touchscreen without the need for typing on a keyboard or using a mouse. Participants' feedback was instrumental in the implementation of key elements of the application including gamification, positive feedback, and background music as means to increase motivation and engagement as well as the use of audio and visual cues as training guides.

The end-product is the proof-of-concept HD-DRUM application that is described in detail in Table 2. HD-DRUM is currently deployed in a randomised controlled trial to assess the feasibility of 2 months of at home HD-DRUM intervention compared to standard medical care control in people with HD. Adopting the IKT framework enabled us to design the HD-DRUM application in a way that maximises the possibility of success of the trial by taking the needs of people with HD into account during the development process.

If successful, the findings of the feasibility study will inform future modifications and refinements of HD-DRUM, as well as clinical outcome measures for fully powered randomised controlled trial to test clinical effectiveness. In the future, HD-DRUM may be able to provide a remotely accessible training tool to help maintain or improve movement and cognition in people with HD without the risk of harmful side-effects. Even a small delay in the onset of symptoms would have direct and significant benefits for the quality of life of people with HD and their families.

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