# Testing the usability and acceptability of the NON-STOP intervention

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| **Study Title** | Evaluating the acceptability and usability of a digital self-management intervention (smartphone application) to support the non-surgical treatment of Perthes' Disease |
| **Study Design** | Mixed methods with observational and qualitative components |
| **Study Participants** | Children with Perthes' Disease and parents/legal guardian(s) (family) of children with Perthes' Disease |
| **Planned Size of Sample** | 30 children with Perthes' Disease (single-arm trial)<br>A subset of 15-21 participants from the single-arm trial will also take part in a nested focus group study:<br>12-15 children with Perthes' Disease<br>3-6 Parents/legal guardian(s) of children with Perthes' Disease |
| **Planned Study Period** | 01/12/2023 – 31/05/2024 (data collection period) |
| **Research Question/Aim(s)** | 1. To test the acceptability of implementing the NON-STOP intervention with children with Perthes' Disease and their families<br>2. Understand the user-experience of children with Perthes' Disease when using the NON-STOP intervention |
| **Sponsor** | University of Leeds |
| **Funder** | HEE/NIHR Clinical Doctoral Research Fellowship. ID: NIHR301228 |
| **Research team** | For the purpose of this study the team is as follows:<br>Chief Investigator (CI) — Anthony Redmond (ACR)<br>Lead Researcher — Adam Galloway (AG)<br>Principal Investigator(s): — Dan Perry (Alder Hey) (DP); Colin Holton (Leeds) (CH); Bree Winger (Sunderland) (BW)<br>Research team — Anthony Redmond (ACR); Suzanne Richards (SR); David Keene (DK); Heidi Siddle (HS) |

# Contents

# 1. Background

## 1.1 What is Perthes' disease?

Perthes' Disease is an idiopathic condition affecting the development of the hip joint in children that involves bone destruction due to disruption of blood supply (avascular necrosis) [1]. During the active disease process, children are often in pain, and typically develop walking difficulties, limiting their activity [2]. Following resolution of the disease process many children are left with significant joint damage, often requiring hip replacement surgery in early adulthood [3, 4]. The negative impacts on health and quality of life for these children and their families can be substantial. Moreover, effects on sleep, play and school attendance may lead to longer-term social and emotional difficulties [5], potentially leading to lifelong consequences on development and limiting life opportunities.

The condition affects around 1 in 1,200 children in the UK. These children are typically aged four to eight years old, with boys affected more commonly at a ratio of 4:1 [6, 7]. Whilst the cause of the avascular necrosis is not known, literature describes an increased incidence in areas of higher levels of socioeconomic deprivation [8]. The condition appears to be much more common in the North of England compared to elsewhere in the world.

The disease burden on children and their families can be compounded by the burden of treatment, which generally involves regular monitoring and various surgical and/or non-surgical treatments. A recent NIHR-funded study reporting the incidence of surgery for Perthes' Disease found that 233/393 (59%) hips received surgical care in the first year of surveillance [9]. As healthcare moves to the 'digital-age', there is more emphasis on providing technology-based healthcare interventions. This has been accelerated by the COVID-19 pandemic and has become common practice in physiotherapy care for children [10]. It is likely there will be a sustained need for digital healthcare to support the traditional management of patients in the future [11].

## 1.2 Why is this research important?

Variation in the management approaches of Perthes' Disease between centres adds uncertainty and worry for families. There are currently no standardised treatment protocols, either nationally or internationally for Perthes' Disease, and limited consensus on treatment strategies. The traditional treatment for Perthes' Disease is non-surgical, with options including orthotic management (e.g. braces and callipers), physical interventions such as strengthening and stretching regimes, walking aids, activity modification, or observation alone [12]. A systematic review, completed by this research team, explored the non-surgical treatment methods for Perthes' Disease and showed no robust evidence to guide practice [7]. Similarly, a multicentre case-review highlighted the variation of care in the UK [13], with one centre where all patients received surgery, and another where just a third received surgery. In one centre all patients were advised to modify their daily activities, whilst this was only advised for a fifth of children in another centre. The reasons for this variation in care are mostly unexplained but may be due to the training/experience of the clinicians rather than any evidence-based benefits for children and their families.

Leading up to this study, we have previously completed a programme of work including a qualitative study exploring the experiences of key stakeholders in relation to care for Perthes' Disease, and a national Delphi study to reach clinical consensus on the non-surgical treatment of Perthes' Disease.

The qualitative study informed the Delphi study by identifying the domains to include, in order to arrive at clinical treatment consensus with clinical experts. It also informed the content of a digital self-management intervention, based on this consensus and that will be used on a smart device, hereafter referred to as the "app" . The app has been developed to assist children in the management of Perthes' Disease and will be tested throughout the proposed research study. The Delphi study refined and guided the clinical content for the app, which has been further developed using Patient and Public Involvement in preparation for testing.

The content of this digital self-management intervention is derived from the recently completed clinical consensus study and the aforementioned studies. As per the findings of said work, an important part of this self-management intervention will be promotion of exercise and physical activity. Physical activity has many benefits, amongst which is improving the strength and stability in the area affected by Perthes' Disease [14]. There are also wider health benefits such as reduced risk of childhood obesity [15]. This is particularly pertinent to children with Perthes' Disease who are often advised to modify symptom-provoking activities, resulting in greater overall sedentary behaviour. Evidence suggests that self-management in adults with long term conditions such as asthma and cardiac disease is more successful if supported by digital interventions that help them to feel more involved in how they manage their care [16]. Whilst the behaviours and motivation for engagement have not been measured in child-health research, a systematic review did summarise health outcomes [17]. The authors suggested that self-management interventions in some conditions (spina bifida and arthritis) lead to better health outcomes.

Self-management has a particularly significant role to play in the treatment of Perthes' Disease, as children typically have long periods of time between hospital appointments for either medical or physiotherapy review. Having a reliable resource to use for information, education and self-management could potentially bridge that gap and supplement existing care. However, there would need to be careful consideration of how any potential novel intervention could be best integrated into the clinical setting. Testing the acceptability of the app (described below) will consider how acceptable the content is to children with Perthes' Disease. The app will supplement, though not replace the need for physiotherapy review which was one of the recommendations from the recent clinical consensus study (not yet published). In this Delphi study, clinical specialists (physiotherapists, clinical nurse specialists and orthopaedic surgeons) achieved clinical consensus that "all children with Perthes' Disease should be offered an initial assessment with a physiotherapist".

This intervention for Perthes' Disease has been developed to fulfil the recommendations of the National Institute for Health and Care Excellence (NICE) evidence standards framework for digital health interventions [18]. Specifically testing the intervention in the domains of 'acceptability with users' and 'effectiveness for preventative behaviour-change or self-manage functions' as part of the single-arm trial.

## 1.3 Have patients and public been involved in this study?

From the outset of this programme of work there has been substantial Patient and Public Involvement (PPI). Children at the NIHR Young Persons Advisory Group at Leeds Children's Hospital and Alder Hey Children's Hospital have been instrumental in the design and development of the study. PPI input has informed the name of the project (NON-STOP: Non-Surgical Treatment Of Perthes') as well as the design of a logo for the project. In the planning and development stages of this study, PPI have helped refine the design of patient facing documents/resources. These include information resources that

participants will receive when considering their involvement. They will also be involved in the design of topic guides for the study. The research team has regular access to the Young Persons Advisory Group as well as a project-specific advisory group for the NON-STOP project. The project advisory group includes two child/family dyads and an adult who has Perthes' Disease and received a range of interventions as a child.

# 2. Research aims and objectives

## 2.1 Aim

a) To test the acceptability and usability of the NON-STOP digital self-management intervention (app) amongst children with Perthes' Disease and their families, and highlight areas for refinement.

## 2.2 Objectives

a) To further develop the NON-STOP app (including training materials for users) amongst a representative sample of children with Perthes' Disease and their families.
b) To explore the acceptability and usability of the app by analysing quantitative information collected by the app during use, and through qualitative focus groups with users.
c) To explore the acceptability of study procedures in preparation for a definitive trial.

# 3. Study design

The over-arching design includes a quantitative single-arm trial exploring app usability and acceptability [19], and a nested qualitative focus group study to explore users' perspectives [20]. This stage of the project is not to evaluate efficacy or effectiveness of the intervention, only to establish and maximise usability and acceptability.

The methods used will be similar to those previously used in qualitative studies including with children [21, 22].

This is the final study in a larger mixed-methods project approved by the National Institute of Health and care Research (NIHR). The overall project aim is to develop and test a digital intervention for children/families to promote self-management of Perthes' Disease by facilitating physical activity. The mixed-methods approach is pragmatic [23, 24] and is underpinned by a person-centred approach intended to test intervention development [25-27]. A mixed-methods approach allows for the synthesis of evidence from both quantitative and qualitative research and in turn supports best practice. The key way in which mixed-methods does this is by allowing researchers to explore the breadth and depth of a problem by combining quantitative data with contextual data which can give more credibility to the results of a study [28]. In context to this mixed-methods study, quantitative data on the usability and acceptability of the app will focus on number of times the app is used, which parts are used, and for how long. The nested qualitative focus group study will use this information to increase the relevance of the findings to all users by, for example, inviting users that have had high and low amounts of app use to discuss reasons they used the app the way that they did.

## 3.1 Health technology assessed: The NON-STOP app

The intervention being assessed is a digital app, which includes a mix of educational resources and exercises. The content for the app is based on best practice recommendations which are derived from a mix of theory and the findings of a recently completed Delphi study, both of which will be explained in this section. It is important to test this new app and assess its acceptability, however it is important to note that the app itself is a method of delivering the care recommended for children with Perthes' Disease rather than a novel way to treat the condition. Children are routinely asked to complete stretching and strengthening exercises and families are provided with educational resources. This NON-STOP app is a novel way to provide this education for children with Perthes' Disease and their families. It is not for monitoring clinical progress or pain by clinical teams.

Children/families who consent to take part in the trial will be given instructions on how to download, register and use the NON-STOP app. They will be given a unique identification number so that the lead researcher can identify the unique user and record data on their app use.

There is limited evidence available around the optimal duration over which a healthcare app should be tested, particularly for children. There are some previous studies that have tested healthcare apps in an adolescent/adult setting and have done this over the course of four weeks [29]. We will test the NON-STOP app over the course of six weeks. This time period is recommended by the independent app-developers who have an extensive history of developing and testing digital health interventions with children [30]. It also aligns with common review points in clinical practice for the assessment of an intervention.

Children/families using the app will have daily alerts/reminders delivered using push notifications on their smart device. The daily reminders will aim to encourage daily use of the app, particularly in the exercise domain. If a child/family has not used the app in 21 days, an alert will be sent to the app developer who will inform the lead researcher (AG). Following this, AG will make a one-off contact the family to offer help/advice with app use and explore any potential reason for non-use. The alert generated by the app to demonstrate non-use will only provide the app development team with the unique user-ID. The lead researcher (AG) will use to identify the participant and contact the participant. More information on the safe-storage of information within this study is provided in section 9.

### 3.1.1 Theoretical approach

Given that the overall project aims are to develop and preliminarily test a digital intervention to promote engagement in exercise and physical activity for these children, the project draws upon the Social Determination Theory (SDT) [31]. SDT is intended to explain how individuals adopt and/or maintain behaviours. This theory states that motivation is linked to the level of three 'psychological needs': autonomy, relatedness and competence [32]. SDT proposes that focus on the importance of intrinsic motivation is beneficial for exercise and physical activity [33]. Previous literature demonstrates success in terms of increased levels of exercise and physical activity when the 'psychological needs' have been addressed. Higher levels of competence and relatedness has proven to influence enjoyment in PE in an educational setting (exercise) where children are in a motivational environment, surrounded by others completing similar tasks.

Whilst SDT is the primary theory underpinning this study, it will be further informed in part by the Social Ecological Model (SEM) of behaviour [34]. Although SDT seeks to explain the factors motivating behavioural change at the level of individuals, SEM has the advantage of placing more explicit emphasis on the importance of environmental factors in which behaviour takes place. This is of

particular importance given that Perthes' Disease is more prevalent in socio-economically deprived children/communities. We will focus on three levels of SEM in this study: the child (applying SDT theory); interpersonal factors (e.g. role of family and peers), and organisational factors (e.g. availability of home and local community environments to support physical activity) which might impact on the self-management of Perthes' Disease. The delivery of the digital self-management intervention will follow the guidance from Borelli et al who, in 2004, outlined a plan for treatment fidelity assessment and implementation [35]. This plan refers to elements of an intervention that can be assessed to measure a key element of fidelity which is treatment integrity. Specific to this NON-STOP app testing; this will be used to measure the degree to which the intervention is delivered as intended.

The benefits of an integrated theoretical approach to intervention design have previously been demonstrated in adolescents [36]. Indeed, it is widely recognised now that theory integration is encouraged, as it can reduce the redundancy between theories and utilise the strengths of specific theories [37]. The behaviours assessed will be around the use of the app as well as the content of the app and training materials (outlined in section below) will be informed by this theory. The qualitative element of this study will involve asking whether participants use the app and what factors influence this. Questions in the focus group will address use-related behaviours. For the app content, we will measure whether the intervention content changes health behaviours in terms of whether it increases outcomes of interest such as physical activity pre and post app-use.

### 3.1.2 Best practice content

The content of the app is informed by the findings of a recent Delphi study completed by this research team. In the absence of robust evidence, this was agreed to be the best format to guide clinical management. The consensus statements include provision of educational resources on Perthes' Disease. Including anatomy of the affected area, typical presentation and disease progress, and relevant research on the topic. The app also includes instructional videos and guidance on stretching and strengthening exercises and the opportunity for users to track their progress using 'activity diaries'.

There are also embedded training materials for users to understand how the app works. Children/carers will be instructed on how to use the app and reiterate how often to complete their exercises as per existing guidance. There are also instructions on the app for when users may need to seek help from their treating team. Reasons for this may be that there is a significant increase in their pain levels or they have specific questions about their care. As discussed, this app is supplementary to existing care and not to replace the care from the clinical team, this will be made clear to all participants using the PIS attached (**PIS APP CHILD & FAMILY)**.

### 3.1.3 Training

When the participant logs in to the app for the first time, they will be taken immediately to an instruction video of how to use the app. The videos will explain in detail how each of the domains works, and various options that are available to the users, for example, children using the app will have a choice of different layouts for activity diaries (superhero theme, race-car theme, unicorn theme, etc.) as well as options for their avatar which they can customise. The app cannot be used without watching this introductory video. The instruction video can be reviewed at any time within the app. All instructional media has been developed with input from key stakeholders including PPI and clinicians in the field of paediatric orthopaedics for quality assurance.

# 4. Sample

## 4.1 Participants and settings

Participants will be children receiving treatment in one of three NHS hospital centres that commonly treat children with Perthes' Disease (Leeds Children's Hospital, Alder Hey Children's Hospital and South Tyneside Hospitals) and their families. Recruitment from different centres is important to ensure the sample is representative of the wider UK patient population. Participants will be recruited from their usual orthopaedic appointments. Children will be sampled purposively to maximise heterogeneity e.g. differing sex, age, duration of living with condition and disease severity [38].

The children/family recruited to the study will be initially identified by a lead-clinician within each centre who is familiar with the study eligibility criteria. This patient population will include children from a range of age groups, and it is possible that participants will be from educational key stage (KS) 1 (aged 5 to 7 years), KS2 (aged 7 to 11 years) and KS3/4 (aged 11 to 16 years). Amongst younger children (KS1/2), data collection will be based on the 'draw and write' technique which has been successful in previous studies with children focussing on physical activity [39, 40]. This method allows the researcher to ask the child to draw pictures based on the subject, in this instance, focussing on their experience of using the NON-STOP app. In turn, the child has the opportunity to share their experiences/feelings through 'storytelling' and opens the dialogue between researcher and other participants within the focus group, a method that has been proven to provoke longer, more meaningful, answers from children.

## 4.2 Eligibility criteria

Participants (including family members) will be eligible for inclusion if the individual affected,

1) Was diagnosed with Perthes' Disease between one and five years ago.
2) Are currently aged between 5 and 16 years old.
3) Have access to a smart device.

Participants (including family members) will be excluded if,

1) They are unable to communicate verbally in English.
2) The child has undergone surgery for Perthes' Disease in the last 6-weeks.

The rationale for excluding children with Perthes' Disease who have had surgery within the last six weeks is that children will be require to complete physical activity in the app. Typically post-operatively children are placed in casts to maintain ROM, or are given weightbearing restrictions and would not be able to complete the exercises/activities advised within the app.

## 4.3 Recruitment

### 4.3.1 Single-arm trial

Recruitment of children for app-testing will take place in the clinical setting, with clinicians in each centre trained regarding the study eligibility criteria and recruitment processes. Once a potential participant is identified, the treating clinician will briefly outline the purpose of the study. If the child/family is interested in hearing more about the study, they will be asked for permission to share their name, email address and phone number with the lead researcher (AG). These details will be passed to AG using secure email (NHS.net). These emails will be securely stored in password protected

files on a University of Leeds server and deleted from the NHS.net email inbox. Permission to pass the contact details of the child/family to the research team does not mean they have agreed to take part in the study. The clinician who obtained permission for the lead researcher to contact the child/family dyad will record this in the child's medical notes.

The lead researcher will then seek consent as outlined in section 4.4 below. Following consent, the lead researcher will provide the child/family with registration information for the app (including a unique study identifier) and link to baseline questionnaire.

Each site will recruit ten children to take part in app testing, recruiting a total of 30 participants. In order to provide a thorough description of the recruitment process, anonymised screening data on number of child approached/agreed will be collected by the recruiting clinicians at each site. In order to investigate the representativeness of the sample the clinicians will provide the lead researcher with age (years), sex and time since diagnosis (months), ethnicity, and reason for not taking part (if applicable) will also be collected. Recruitment will be monitored based on the characteristics outline above, which are based on the prevalent characteristics in the patient population recorded in the literature, as well as known from the research team's clinical experience.

### 4.3.2 Nested focus group study

Children/families will be asked to indicate if they are willing to be approached to take part in a focus group as part of the baseline questionnaire. Those who express an interest will be contacted by the lead researcher approximately two-weeks prior to the end of the six-week app-testing data collection period. The consent process is outlined in section 4.4. From the 30 trial participants, 4-5 child/family pairs (dyads) will be recruited to take part in a focus group at each clinical site (12-15 participants in total). This group size is selected based on guidance in the literature around how to optimise focus group engagement with children [20]. In the event of not achieving this sample size, a smaller focus group will be used and understandable limitations discussed in the eventual scientific write-up. The team are, however, confident of achieving this sample size based on recruitment rates in the recently completed qualitative study.

Kennedy et al in 2001 provided some guidance on when to conduct focus groups with young children (age 6-12 years) which is relevant to this patient population [41]. Specifically, that focus groups are effective when looking to evaluate an intervention and provide rich and meaningful responses from children, which may not always possible in more structured qualitative data collection methods such as interviews. The authors outlined key mechanisms to encourage interaction and engagement in focus groups with children that will be adopted here, including: increasing the comfort of children by ensuring there was a peer audience (i.e., children and parents not involved in the same focus group), and to adopt language that is appropriate and applicable to children.

## 4.4 Consent/assent

Informed consent will be gained for all participants (parents/legal guardians and children). Given the involvement of children under 16 in the study, a parent/legal guardian will be required to provide consent for their child and the child will provide assent.

### 4.4.1 Single-arm trial

Following identification of an eligible child/family, the lead researcher will send them an email with the participant information pack that includes an information sheet for the parent and a separate age-specific information sheet for the child **(PIS APP CHILD & FAMILY)**. In line with current HRA guidance, families will have at least 24 hours to consider the information before the lead researcher contacts

them to discuss study participation. However, if a family responds sooner than this, then the team will try to accommodate this [42]. If the family has not responded within one week, a second email (same content as first) and/or text message will be sent, there will be no further attempt to contact after this.

A proportionate consent process will be adopted, based on NHS Health Research Authority guidance and given the low risk of the data that will be collected from this research [42]. This method for consent was deemed appropriate for the authors' recent qualitative interview study involving children with Perthes' Disease, their families, and clinicians (REC reference: 21/WS/0138). On contacting the participant, the parent/legal guardian will be asked to provide informed consent electronically via email. The participant will be asked to reply to the email and include the 'statement of agreement' that can be copied from the original email to read as follows:

> *'I have studied the information provided in the participant information pack and understand what will be required of my child/me during this study. I give consent to the use of any information gathered during this study for the purposes outlined by the research team. I also understand that my/my child's participation is voluntary, and I am free to withdraw from this study, unchallenged, at any time.'*

On return of this statement of agreement, baseline data collection can begin (see section 6). After providing consent to taking part in the trial, each participant completes an online questionnaire using Online Surveys [43]. This is primarily to provide some demographic information and baseline measurements. They will also at be asked to indicate whether they are willing to be contacted about taking part in a focus group to discuss their experiences of using the NON-STOP app at the end of the six-week trial. There will be a "yes" and "no" option and a footnote that explains that their answer does not affect their eligibility to take part in the trial.

### 4.4.2 Nested focus group study

For trial participants that indicated a willingness to take part in the focus group study, an invite will be sent outlining the nested qualitative study. The lead researcher will contact participants who expressed an interest in taking part in a focus group and provide the study information (**PIS QUAL CHILD & FAMILY**). The same proportionate email-based consent process described for the trial participants will used for focus group participants.

On completion of consent for child and/or family participant, the focus group will be arranged. At the start of the focus group, the child and family will be reminded that the process can be stopped at any time. To confirm their agreement to take part, and to gain verbal assent from the child, each participant (child or family member) will be asked at the start of their focus group if they are happy to continue and reminded that at any point they can withdraw.

It will be made clear to the participants that their involvement in the study (focus group and/or trial) will remain confidential from their treating clinical team. Inclusion in this study will in no way affect their clinical care now or in the future.

# 5. Data collection

## 5.1 Single-arm trial

Baseline data entry includes participant demographics and patient reported outcomes including the PROMIS Mobility [44] and Children's Physical Activity Questionnaire (CPAQ) [45, 46]. The PROMIS Mobility is a validated outcome measure for children with Perthes' Disease [44] and will be completed before and after the app-testing period. Previous studies have demonstrated that it can be completed by children eight years-old and above, and that for children younger than eight years-old, an adult proxy is recommended [47, 48]. PROMIS is scored using a T-score in which the total raw score of the measure is converted and a higher score indicates more of the concepted being measured. For example, if measuring mobility, a higher T-score suggests more mobility than a low score [47].

CPAQ is a questionnaire that reports the physical and sedentary activity of a child and has been validated and implemented in studies with children as young as four years-old [46, 49]. CPAQ is generally reported in minutes per day for each activity [49] and will be reported pre and post intervention.

Children/families will be able to record pain if it occurs during the app testing on their activity diary. They will be able to use the Wong-Baker FACES method of reporting pain. This method is commonly used in paediatric practice both in a clinical and research setting [50, 51]. The child selects a face that represents low to high levels of pain on a scale of 0-10 (shown below in Figure 1) and has a proven correlation to the Visual Analogue Scale for pain [51]. It is worth reiterating that the collection of pain information is purely for the research team after the app-testing period. Information collected during the period will not be monitored nor used. There will be clear instructions for users advising them to contact their existing clinical care team if they have specific questions/concerns about their pain levels.

*Figure 1 – Wong-Baker FACES scale*


At the end of the 6-week period, participants will complete the same outcome measures as pre-testing (PROMIS Mobility and CPAQ) as well as the Health-ITUES outcome measure [52]. Health-ITUES is a validated measure used to assess usability of a digital tool [53, 54]. This 20-question, customisable questionnaire is used to assess usability and acceptability in four domains [55]. The four domains are impact, perceived usefulness, perceived ease of use, and user control. All of which relate well to the aims and objectives of this study. The items within the questionnaire are rated on a Likert-scale from strongly disagree (1) to strongly agree (5). Each item is weighted equally and a mean score from each domain is calculated.

All questionnaire-based assessments will be completed using an online form using Online Surveys, which has been used in a recent study as part of the over-arching doctoral programme by the lead

researcher (AG). Participants cannot progress without completing all relevant sections of the questionnaires. Setting up the assessments in this way removes the risk of missing data from assessments. The table below shows an overview of the assessments during the study and when they will take place.

Table 1 – Showing content and timing of assessments during single-arm trial

| **Data collected** | **Baseline** | **6 weeks** |
|---|---|---|
| Demographic info | X | |
| PROMIS Mobility | X | X |
| CPAQ | X | X |
| Health ITUES | | X |

The single-arm trial will also include process measures from the app that will be collected and assigned to the participant identification number. This information is collected via a database developed by the app-development company, HMA. Process data will be collected as follows:

- Number of app log ins (total and each day)
- Which pages/sections of app accessed during app-use (each log in, list of pages accessed and for how long)
- Time of day accessed (measured for each log in for each participant, to assess trends in use i.e., before or after school, weekends, etc.)

Initial engagement:

- Do they register to use (yes/no)
- Did they complete the training package (watch the video)

## 5.2 Nested focus group study

Once a date/time suitable for participants has been assigned for the focus groups for either child/family dyad, the focus groups will take place with an independent facilitator and the lead researcher (AG) as note-taker. The facilitator is a children's specialist physiotherapist who has experience of communicating with children with Perthes' Disease and their families. They will facilitate the discussion within the focus group whilst the lead researcher (AG) observes and takes notes to accompany the transcripts to ensure any inaccuracies or failures in recording. Data collection will follow the relevant topic guides/schedules for the participant. Focus groups will take place on the same date, but there will be a separate focus group for children and family members. There are provisions put in place for childcare/supervision of the children whilst the family-participants take part in the focus groups.

These focus groups will follow a topic guide (detailed below) and will take place face to face at each of the three clinical sites used for recruitment. A non-clinical setting has already been identified in Leeds and Alder Hey from previous PPI activities, for the remaining site (South Tyneside), a non-clinical setting will be identified in order to provide a setting that promotes conversation within the focus groups.

All focus groups will be recorded on an encrypted audio recording device. Topic guides will be developed using a combination of sources. Some examples of topics to be included are detailed in the section above such as use of the app, when and where the app gets implemented and different

elements of the app (exercises, activity logs and information section). The guides will also be guided by input from PPI group outlined previously as well as the experience of the research team.

*Figure 2a – Single-arm trial process*

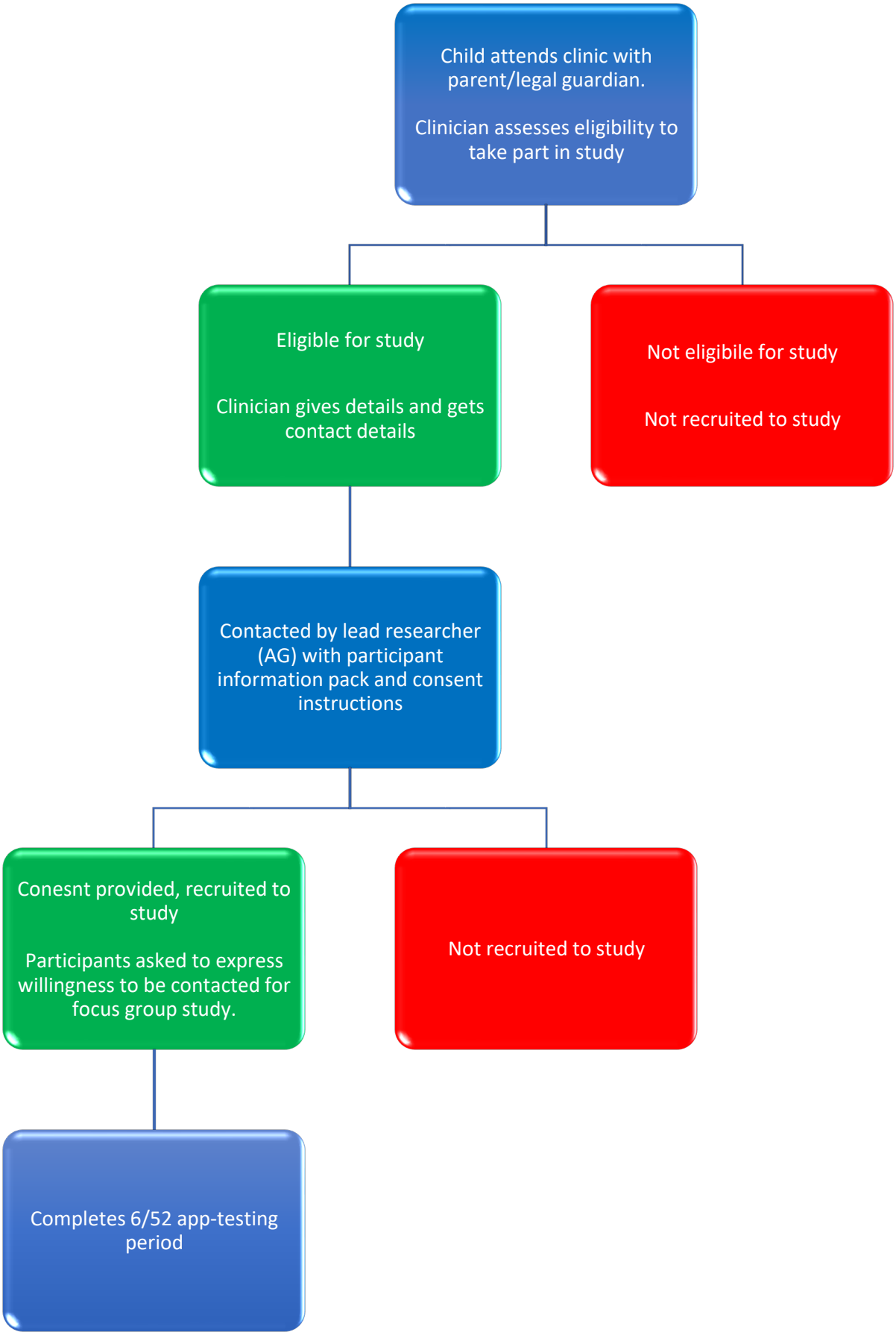
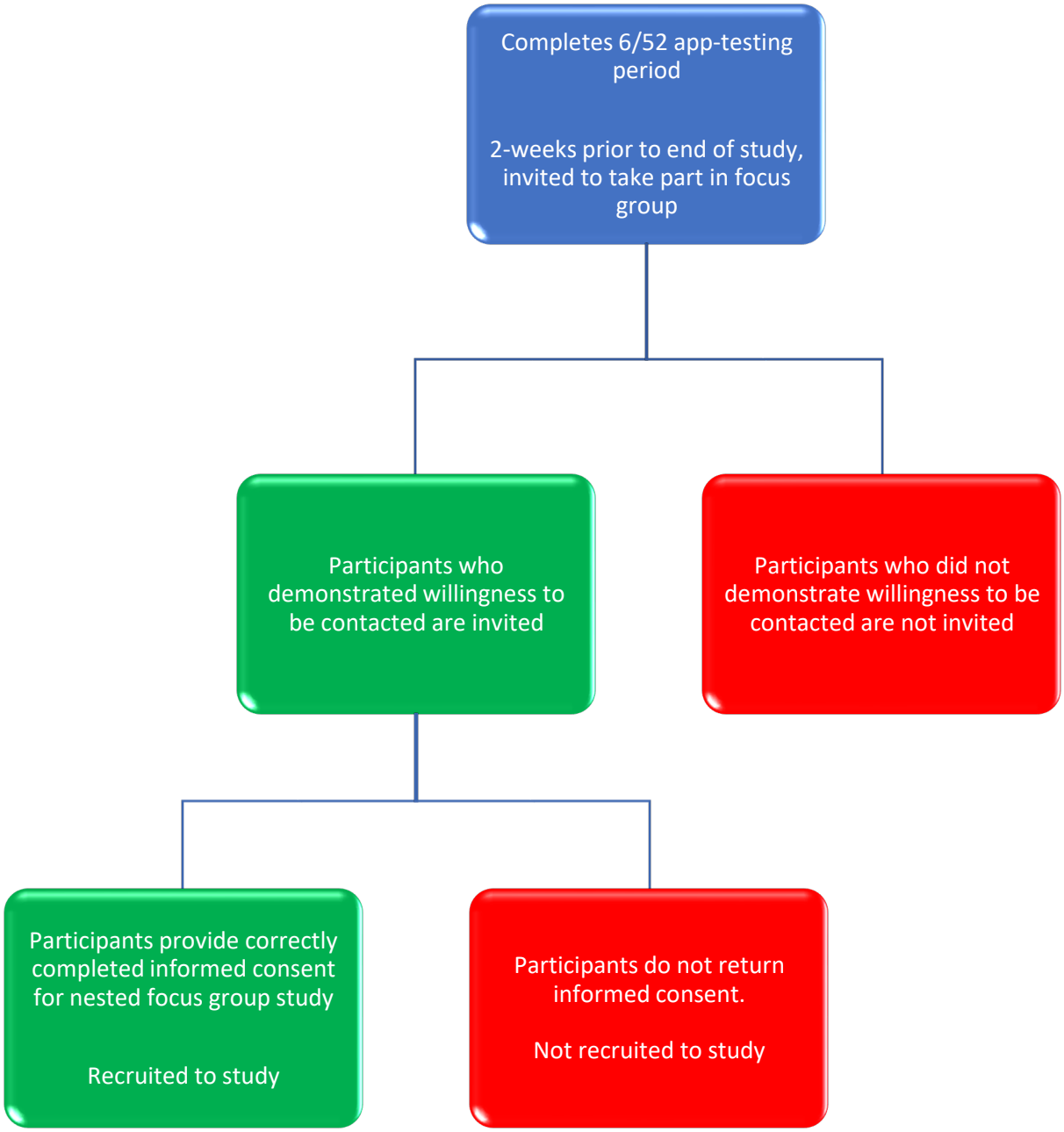


*Figure 2b – Nested focus group process*



# 6. Data analysis

## 6.1 Single-arm trial

Descriptive statistics will be used to summarise participant characteristics (baseline), process measures (post-intervention follow-up) and outcome measures (baseline and follow-up). There will be no inferential statistics used in this acceptability and usability study.

To address methodological uncertainties prior to a definitive trial, descriptive statistics will be used to describe participant recruitment, retention and attrition rates. Clinicians collecting contact details will provide number of participants approached. From this it will be possible to provide the number of participants who agreed/did not agree to being contacted about the study. The number of participants who then accepted/declined study participation will be collected. Attrition over the course of the six-week testing period will be described.

The process measures described in the data collection section will be analysed descriptively, with an overview of trends in things such as number of log ins for each participant as well as the cohort (which would be presented as a mean with range). Most-visited pages will be calculated for each participant and as a cohort. To analyse the time of the day accessed, a time-threshold will be used to measure whether participants use the app in the morning, afternoon, or evening (after 16:00 to account for use outside of school hours). We will report this as a most commonly used time for participant as well as for the whole cohort.

The results of PROMIS Mobility are presented and translated into a T-score metric, with a mean of 50 and standard deviation (SD) of 10 in the referent population [56]. The T-score for participants pre- and post-app testing will be provided for participants. For CPAQ, as described in data collection section, the minutes per day per activity will be provided. This will be described for the total cohort both pre- and post-app testing. The HealthITUES measure is rated on a Likert-scale of strongly disagree (1) to strongly agree (5) across four domains. A mean score in each domain will be calculated. There are no validation studies conducted in children with Perthes' Disease but available literatures suggests an optimal cut-off point of 4.32 defines usability [57].

## 6.2 Nested focus group study

Anonymised focus group transcripts will be exported into NVivo software and analysed following a Framework approach which will be underpinned by the behaviour theory (SDT and SEM). Consistent with a framework approach, some coding will be deductive, following the structure and questions included on the topic guide (including the theoretical underpinnings). An inductive approach [58] will also be used however, to identify concepts emerging directly from the data. Salient themes and concepts will be identified through thematic coding [59]. Transcripts will be coded iteratively, with preliminary codes revised in light of coding of subsequent transcripts and applied to all. The first focus group will be coded independently by a member of the research team (SR) checking for agreement on emerging codes. Inconsistencies in coding will be discussed, and agreement reached on the subsequent coding which will be reapplied to earlier transcripts, any disagreements will be settled using a third coder.

Positional reflexivity (i.e. reflecting on how this might affect the analysis) will be considered during these discussions [60]. This is based on the lead researcher's existing role as a children's physiotherapist with a special interest in Perthes' disease. This will be balanced by the involvement of the wider research team. For example, the second coder in this instance, SR, is not a physiotherapist

and has no prior knowledge of the condition, nor it's patient population. Study participants will be sent a summary of the main findings and given the opportunity to comment on our interpretation of data.

# 7. Safety reporting

The research team have not identified any potential for serious adverse events (SAE) given the nature of the study. However, in the interest of maintaining the duty of care to the patients, any safeguarding issues that may arise when conducting the focus groups with child/family dyads will be escalated to the PI at the local site who can then follow their local policy. This process will be explained to the PIs in the study set up and any issues/queries can be addressed here however given the close working of each of the PIs there are no anticipated issues regarding escalation of any particular safeguarding concerns.

## 7.1 Indemnity

The University, when acting as Sponsor, has insurance cover in force, which meets claims against it and where those claims arise from the Universities own negligence in its role and activities relating to the study (and which is subject to the terms, conditions and exceptions of the relevant policy). Clinical negligence indemnification will rest with the participating NHS Trust under standard NHS arrangements.

# 8. Approvals/ethical consideration

NHS ethics and HRA approval will be sought for the purpose of this study given the involvement of NHS patients as well as family members. This is in line with the most recent regulatory changes to doctoral student research [61]. Approval from University of Leeds, as sponsor, will be obtained as part of the approval process. Approval from additional sites research and development departments will also be obtained.

There is a certain degree of inconvenience to consider with regards to participant time. This has been considered in the methods and minimised where possible, for example, child/family dyads will be recruited from pre-existing orthopaedic appointments as per their usual care. If needed for face-to-face attendance at focus groups, there is a provision for childcare cover and an allowance for payment for participant time. A provision for an allowance as payment for participant time has been made. A rate of £50 per participant has been funded by NIHR for this study.

## 8.1 End of study and deviations from protocol

Definition of end of study will be confirmed as the end of data collection, which as described, will be 31/05/2024. As per the guidance from the Standard Operating Procedures for Research Ethics Committees, any deviations from this protocol and/or serious breaches to the protocol will be reported to the sponsor within one working day of research team awareness using governance-ethics@leeds.ac.uk and REC within the required timelines [62].

# 9. Data management

All members of the research team will comply with the requirements of the General Data Protection Regulation (GDPR) and Data Protection Act 2018. This includes the collection, storage, processing and disclosure of personal information, they will uphold the GDPR's and Act's core principles. All information collected during the course of the study will be kept strictly confidential, unless there are any legal or safeguarding concerns which require confidentiality to be breached. If any legal or safeguarding concerns do arise, they will be managed in line with the University of Leeds Research Ethics Policy. This will include considering the situation on an individual basis and asking the University Secretary for guidance if required. Given the nature of the study, it is very unlikely that it will be necessary to breach confidentiality.

The data collected in the questionnaires via Online Surveys will be exported using the existing online function which exports results to Microsoft Excel format. This will be stored on the University of Leeds servers also, in password-protected files. It is anticipated that only the lead researcher (AG) will need to access this, so the password will not be shared with the research team unless deemed necessary, for example in the event of AG no longer working in the institution before the 10 years of data holding has passed. After exporting from Online Surveys, the online account will be erased.

In line with GDPR guidelines, all participant information, including proof of informed consent, will be anonymised using a code-link document and stored in password-protected files on University of Leeds servers that only the research team will have access to. The code-link document will be password-protected and hold the details of participant identification number and their name, this file will be kept for 10 years in the secure files described (University of Leeds servers). The contact details of participants will be stored until the end of the study, at which point the participants will receive summaries of the results, after which their details will be destroyed.

As outlined in the data collection section above, the app-use data will be exported by the app-development company (HMA). The information will not be identifiable to HMA because each user will have a unique identification number that the app development team will not have access to.

Digital recordings of focus groups will be electronically transcribed using a transcription service. The recordings and transcripts of these focus groups will be stored in password-protected files University of Leeds servers for at least ten years in accordance with GCP and MRC guidelines [63]. Given their particular specificity to this study as well as the potential to hold patient-identifiable information, they will not be stored in a repository. Other than in the consent provided in email (statement of agreement), participants will be referred to by their study participant number/code, not by name.

## 10. Dissemination

On completion of this study, a report will be created and filed with the funder (NIHR) using the appropriate methods set out in the fellowship agreement. The study will be written up as a scientific journal article and published in relevant, peer-reviewed journals as well as presented at relevant conferences. There will be a public dissemination plan to share the findings as well as process using social media, with careful measures taken to ensure the confidentiality of all participants. All participants will receive summaries of their input and findings of the study once available.

To ensure an effective dissemination of the results of this study, digital platforms will be utilised to share the findings with relevant parties. For example, videos with infographics outlining the

anonymised answers children/families who took part in focus groups provided and what insight this has given us into their experiences of care. These videos/digital information resources will be shared on social media pages as well as platforms via an existing relationship with STEPS Worldwide. This charity supports children with Perthes' disease and their families, they have a strong presence on all social media platforms and regularly seek new evidence to share with their followers.

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