Development of a digital self-management intervention for the Non-Surgical Treatment of Perthes' Disease: The NON-STOP app

Adam Marco Galloway

Submitted in accordance with the requirements for the degree of Doctor of Philosophy

The University of Leeds, School of Medicine

December 2024

Intellectual property and publication statements

The researcher confirms that the work submitted is his own, except where work which has formed part of jointly authored publications has been included. The contribution of the researcher and other authors to this work has been explicitly indicated below. The researcher confirms that appropriate credit has been given within the thesis where reference has been made to the work of others.

Chapter three includes work from the following published paper:

> Galloway, A. M., Pini, S., Holton, C., Perry, D. C., Redmond, A., Siddle, H. J., & Richards, S. (2023). "Waiting for the best day of your life". A qualitative interview study of patients' and clinicians' experiences of Perthes' Disease. *Bone & Joint Open*, *4*(10), 735-741. https://doi.org/10.1302/2633-1462.410.BJO-2023-0108.R1

Chapter four includes work from the following published paper:

> Galloway, A. M., Keene, D. J., Anderson, A., Holton, C., Redmond, A. C., Siddle, H. J., Richards, S., & Perry, D. C. (2024). Clinical consensus recommendations for the non-surgical treatment of children with Perthes' Disease in the UK. *The Bone & Joint Journal*, *106-B*(5), 501-507. https://doi.org/10.1302/0301-620X.106B5.BJJ-2023-1283.R1

Chapter five includes work from the following paper, which at the time of submission has been accepted for publication in August 2025:

> Galloway, A. M., Anderson, A., Casimir, E., Holton, C., Keene, D. J., Redmond, A. C., Siddle, H. J, Richards, S., & Perry, D. C. (2024). From theory to practice: Insights into intervention development of the NON-STOP app for children with Perthes' Disease. *Bone & Joint Open.*

Chapter six includes work from a paper that at the time of submission is under review.

The details from the papers above are included in the thesis chapters from Chapter three onwards. The findings are integrated into subsequent chapters in line with the methodology.

The researcher is the first and corresponding author on all of the above papers. The researcher led all aspects of the work reported including study conception, study design, data collection, data analysis, data interpretation, drafting the manuscripts and responding to reviewers' comments on the papers.

The supervisory team contributed to the revision of the papers, and approved final versions. Their contribution to specific aspects of the papers are described below:

Chapter three: Qualitative study

SP: study design and data interpretation
CH: study conception and study design
DCP: study conception and study design
ACR: study conception and study design
HJS: study conception and study design
SHR: study conception, study design and data interpretation

Chapter four: Clinical consensus recommendation study

DK: study design and data interpretation
AA: study conception and study design
CH: study conception and study design
ACR: study conception, study design and data interpretation
HJS: study conception, study design and data interpretation
SHR: study conception, study design and data interpretation
DCP: study conception, study design and data interpretation

Chapter five: Development of the NON-STOP app

DK: study design and data interpretation
CH: study conception, study design
ACR: study conception, study design and data interpretation
HJS: study conception, study design and data interpretation
SHR: study conception, study design and data interpretation
DCP: study conception, study design and data interpretation

This copy has been supplied on the understanding that it is copyright material and that no quotation from this thesis may be published without proper acknowledgement.

The right of Adam Marco Galloway to be identified as author of this work has been asserted by Adam Marco Galloway in accordance with the Copyright, Designs and Patents Act 1988.

Acknowledgements

The acknowledgments section of this thesis has the highest version count of the entire PhD. I started writing it the second I was awarded the fellowship. One page will never do it justice, but in an attempt to give credit to those who I could not have done this without, here goes.

My supervisors. My “dream team”. Professor Anthony Redmond, Professor Heidi Siddle, Professor Suzanne Richards, Professor Daniel Perry, Mr Colin Holton and Dr David Keene. Over the course of a four-year PhD, I have had supervision whilst swimming in Roundhay lake at 06:00, at open-mic nights in Garforth, and at the Grand Ole Opry in Nashville…I did, however, avoid any hobby-based supervision sessions with the Blackbelt-Prof! Wise words, reassurance and guidance has resulted in something I could have never hoped for, a doctorate. Thank you.

The culture that I have been surrounded by is something I look forward to fostering further in my career, but to those that have made this all the more special, I thank you too. The FASTER team in Leeds is a special team to be a part of, and I am grateful to call myself a member. I must thank the NIHR, without whom this project would not have been completed, and my career as a clinical academic would not have developed as it has. I am proud to be a part, and hope to contribute to the excellent work that the NIHR supports. Additionally, to collaborate with a company like HMA who produce the level of quality that they do whilst making you feel like family has been a highlight, and I hope for more collaboration in the future. Bringing the NON-STOP app to life has been a fantastic experience.

I owe a great deal of thanks to friends and family who have listened to me harp on about Perthes’ Disease, and politely smiled and nodded as I witter about how I might one day contribute to the knowledge somehow. No one has endured this Perthes’ geek like Laura. Testing out frog-jumps, to having hip ROM assessed at random times to appease me. Thank you for your patience and unwavering support.

I am so proud of the PPI and project advisory group members who guided this project from the first YPAG meeting in Liverpool, where Bobby the Bone was born, through to seeing the Bobby pedal on a bike as the NON-STOP app loaded up. Loredana, as chair you were excellent, and I cannot thank you enough for the introduction to STEPS. Michelle, Bev, Fred, Tammy and of course, Teddy! Your input to this project has been invaluable, and I am so proud to have had you contribute to this project.

Finally, the participants, the children and families who gave up their time to come to extra events, take part in interviews and the clinicians who spent time filling in ‘just another survey’. None of this work would have been possible without you. Thank you.

Abstract

Perthes' Disease is an idiopathic avascular necrosis of the developing femoral head that causes joint deformity and significant impact on a child's physical, mental and social health. In the children's orthopaedic community, Perthes' Disease is a top priority for further research. From a non-surgical perspective, there is widespread variation of care and there is no robust evidence to support any specific non-surgical approach, but it often includes physiotherapy and self-management. Patient and Public Involvement work in preparation for this PhD identified that a digital self-management intervention (an app) was an appropriate way to support the non-surgical treatment of Perthes' Disease.

A mix of methods were used to deliver a digital self-management intervention for the non-surgical treatment of Perthes' Disease (the NON-STOP app) and conduct feasibility testing in preparation for further evaluation. A clinical consensus study amongst children's orthopaedic specialists provided recommendations for the non-surgical treatment of Perthes' Disease. The findings of this study provided clinical content that was integrated into the NON-STOP app such as selection of exercises for children to complete and topics for the educational component within the app. During qualitative interviews with key stakeholders, including children with Perthes' Disease, their families and the clinicians who care for them, experiences of existing care were shared. The NON-STOP app was created in collaboration with app developers and PPI input. Children and their families highlighted important features and aspects to improve engagement with the app. These ideas, such as rewards for the avatar 'Bobby the Bone' after using the app, were integrated into the design and development of the NON-STOP app. The app was tested in a mixed-methods feasibility study to explore its usability and acceptability. Children with Perthes' Disease and their families engaged with the NON-STOP app and found it usable. Those who took part in the qualitative element of the study provided insight as to how engagement could be improved and sustained. Suggestions were made regarding intervention dosage and improved rewards.

Due to the methods employed, and most importantly, due to the involvement of relevant key stakeholders considered at every stage, it has been possible to design, develop and preliminarily test the NON-STOP app. It has been refined based on the findings of this project, with updates aimed to optimise engagement. Following this, it has now been integrated into a large, NIHR-funded, multi-centre, randomised clinical trial to compare the surgical and non-surgical treatment of Perthes' Disease.

Contents

List of tables

List of figures

Abbreviations

AMSTAR – Assessing the Methodological Quality of Systematic Reviews

APCP – Association of Paediatric Chartered Physiotherapists

ASD – Autistic Spectrum Disorder

BCT – Behaviour Change Techniques

BCW – Behaviour Change Wheel

BOSS – British Orthopaedic Surgery Surveillance

BSCOS – British Society for Children's Orthopaedic Surgery

CNS – Clinical Nurse Specialist

COM-B – Capability, Opportunity, Motivation and Behaviour

COS – Core Outcome Set

CPAQ – Children's Physical Activity Questionnaire

CTEV – Congenital Talipes Equinovarus

DHT – Digital Health Technology

DTAC – Digital Technology Assessment Criteria

ESF – Evidence Standards Framework

Health ITUES – Health Information Technology Usability Evaluation Scale

HRA – Health Research Authority

HRQoL – Health-related Quality of Life

IPSG – International Perthes' Study Group

IQR – Interquartile Range

IRT – Item response theory

MDT – Multi-disciplinary Team

MRC – Medical Research Council

MRI – Magnetic Resonance Imaging

NGT – Nominal Group Technique

NICE – National Institute for Health and Care Excellence

NIHR – National Institute for Health and Care Research

OMERACT – Outcome Measures in Rheumatology

PAG – Project Advisory Group

PI – Principal Investigator

PiiAF – Public Involvement Impact Assessment Framework

PPI – Patient and Public Involvement

PROMIS – Patient Reported Outcomes Measurement Information System

PROMs – Patient-Reported Outcome Measures

RAM – RAND University of California Los Angeles appropriateness Method

RE-AIM – Reach, Effectiveness, Adoption, Implementation and Maintenance

REC – Research Ethics Committee

ROM – Range of motion

SAE – Serious Adverse Events

SCIENCE – Surgery or Casts for Injuries of the Epicondyle in Children's Elbows

SD – Standard Deviation

SDT – Self-determination theory

SEM – Socio-ecological model

SMO – Systematic Methods Overview

T-CaST – Theory, Model and Framework Comparison and Selection Tool

THR – Total Hip Replacement

TIDieR – Template for Intervention Description and Replication

YPAG – Young Persons Advisory Group

Chapter 1 – Introduction to thesis

1.1 Introduction

This first chapter of a thesis completed as part of a doctoral programme of work focused on the development of a digital self-management intervention for the non-surgical treatment of Perthes' Disease. In this chapter, a summary of Perthes' Disease and the rationale for completing this project is provided as well as the aims and objectives. Finally, a summary of subsequent chapters is provided to demonstrate the structure of the thesis.

1.2 Perthes' Disease

In 1910 Arthur Legg, Jacques Calvé and Georg Perthes published the first descriptions of a child with, as Legg described "an obscure affection of the hip-joint" [1-3]. As a result, the condition was termed Legg-Calvé-Perthes' Disease, most commonly shortened to Perthes' Disease. Over a centruy has passed, and a large degree of uncertainty remains regarding optimal management of this rare condition. This section presents an overview of the condition and the development of the project.

1.2.1 What is Perthes' Disease?

Perthes' Disease is an idiopathic avascular necrosis of the developing femoral head that most commonly affects children aged between four- and eight-years old, with boys affected at least four times more than girls [4]. A disruption in the blood supply to the femoral epiphysis occurs in the early stages of the condition and results in varying degrees of collapse at the femoral head [5]. The true aetiology is unknown however the pathophysiology of the condition is well described through several stages that occur during the disease process. Typically, this process takes between two and five years [4]. Firstly comes the initial stage in which the blood supply is disrupted, which leads to sclerosis and flattening of the femoral head. In the latter

stages of the condition, the fragmentation and reossification stages, the necrotic bone is reabsorbed by the body and new bone is produced, and remodelled until the healed stage is reached [6]. These stages of the condition, originally described by Waldenström have since been modified by Jospeh et al [7]. The modified classification includes an 'early' and 'late' section of the initial, fragmentation and reossification stages to allow for more detailed staging [7, 8]. The stages of Perthes' Disease can be seen in the series of radiographs in Figure 1.1 [9].

Figure 1.1 – The stages of Perthes' Disease using Waldenström classification*

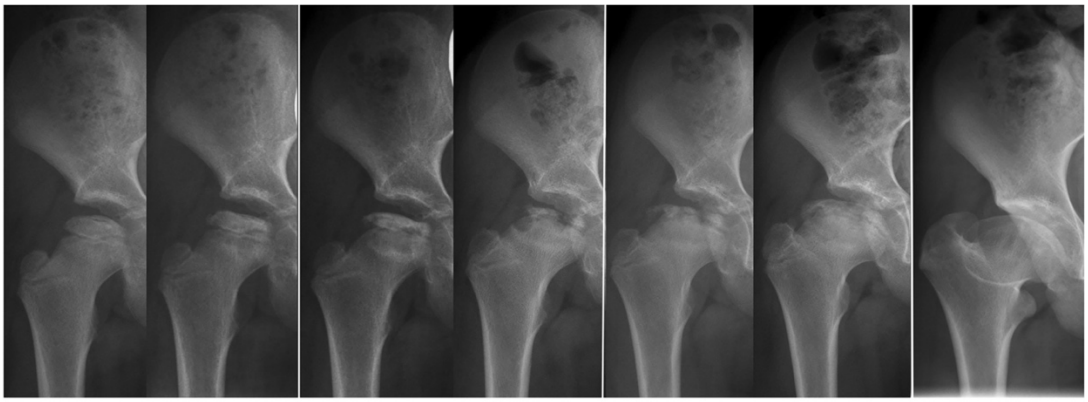


Image from* Varela-García, M., et al., *A retrospective study describing the acetabular consequences of Legg- Calve-Perthes' Disease.* BMC Musculoskeletal Disorders, 2024. **25(1): p. 753.

The shape of the femoral head at the healed stage is dictated by how well the femoral head (despite any change in shape) remains within the confines of the acetabulum of the pelvis during the first three stages of the condition. This concept is called containment, and is discussed in more detail in Chapter two, along with how this is achieved. The change in the shape of the femoral head over the course of the disease process often causes significant implications for the child. These include limited range of motion (ROM), pain, mobility limitations and a reduction in quality of life [10]. The impact on quality of life has been measured using the same outcome measure for Perthes' Disease as in childhood cancer [11]. The participants reported scores that demonstrated similar, and at times, lower scores for quality of life for Perthes' Disease when compared with childhood cancer, demonstrating the impact of the condition on quality of life.

Children will typically present with a limp, and pain in the hip, although this can present as pain in the knee. There is often a restriction in ROM which is most commonly in abduction and internal rotation of the hip. Diagnosis is generally confirmed using plain radiographs of the hip/pelvis. Other diagnostic investigations include Magnetic Resonance Imaging (MRI) [12].

Radiographs are used once the disease reaches the healed stage to predict the long term outcome, which is based on the final shape of the hip. This is classified according to the Stulberg classification [13], which offers clinicians the ability to quantify the degree of deformity and use this to predict the potential onset of degenerative joint disease such as osteoarthritis.

1.2.2 Epidemiology

The UK has the highest incidence of Perthes' Disease in the world, with an annual incidence as high as 19 per 100,000 0- to 14-year olds per year in some areas of the UK [14]. It most commonly affects boys aged between four and eight years old, with males affected around four times more commonly than females [4]. There is a significant variation in the rates of Perthes' Disease depending on location. This geographical variation exists internationally, with regions close to the equator having lower incidence than those in Northern Europe, which has the highest rates worldwide [15]. The geographical variation also exists on a smaller, more local scale [16]. For instance, in the UK, there are much lower rates of Perthes' Disease in London in the south of the UK compared to the North of England and Scotland. The rates in Scotland, in a study in 2012, demonstrated over twice as many cases reported [14]. There is a marked socioeconomic gradient associated with Perthes' Disease, with the most deprived areas in the UK demonstrating rates four times greater than the most affluent areas.

The aetiology of Perthes' Disease is unknown, however there are certain risk factors that have been identified which suggest that the aetiology could be environmental given the link with socioeconomic deprivation. There are associations with certain factors such as exposure to cigarette smoke [17, 18], low birth weight and birth length

[19, 20], atypical stature [21, 22] and other abnormalities such as behavioural issues like hyperactivity [23, 24]. Whilst these factors appear to be important, particularly in such determinants as socioeconomic deprivation, the mechanisms remain unclear.

In recent years, the epidemiology of Perthes' Disease has been well described in the literature, with a large surveillance study of children with Perthes' Disease called the British Orthopaedic Surgery Surveillance (BOSS) Study. This study produced a mass of updated information on the condition within the UK [25]. The data collected in the BOSS study confirmed the findings from 2011 which reported a decline in rates of Perthes' Disease in the UK. That being said, there is still a significant volume of cases in the UK, with data on almost 400 incident cases of Perthes' Disease collected from hospitals in the UK over a two-year period.

1.2.3 Current management of Perthes' Disease in the UK

A particular focus on the literature surrounding the management of Perthes' Disease is provided in Chapter two. In order to provide an overview of how Perthes' Disease is managed, a summary is provided here with reference to the latest available evidence.

In 2007 there was a survey amongst members of the British Society for Children's Orthopaedic Surgery (BSCOS) which collected information about how members managed children with Perthes' Disease [26]. In this study, respondents (n=88) demonstrated that 90% were advised to complete physiotherapy. There was no detail as to what physiotherapy intervention entailed, however it is generally accepted that maintaining ROM and strength of the hip are common practice in physiotherapy for children with Perthes' Disease. This is often done with regular input in clinic settings, hydrotherapy and prescription of home exercise programmes to complete. This approach is often combined with advice regarding activity modification such as reducing high-impact activities, which it is theorised exacerbates deformity in the active stages of the condition [27].

In 2020, working as the lead researcher (referred to subsequently as 'the researcher'), I was able to further demonstrate the variation in care for children with

Perthes' Disease in the UK [28]. In this case review centres in the UK provided information regarding what input children had received in their respective hospital. There were variations in which children received physiotherapy or not, variations in which children were advised to modify activities and whether children received surgery or not. The BOSS study in 2022 delivered further information regarding the management of children with Perthes' Disease in the UK [25]. In the two-year period, 33% (n=117) children were treated using surgery, 67% (n=207) were referred to physiotherapy. The factors that influenced the choices made by surgeons were often clinician-specific rather than consistent patient- or disease-specific factors. The results of the BOSS study demonstrated that age, sex and radiological collapse were identified as predictors of poor radiological outcome, which is in keeping with the literature [29, 30]. Despite the focus that many surgeons put on hip 'stiffness', this did not affect radiographic outcomes. Finally, the BOSS study demonstrated that when containment surgery was carried out, there was no evidence to suggest that it had any bearing on the radiographic outcome. It is reasonable to suggest that here that decisions were based on clinical intuition rather than established prognostic indicators.

1.2.4 Rationale for this programme of work

In 2018, a James Lind Alliance Priority Setting Partnership exercise took place for elective lower limb conditions in children's orthopaedics [31, 32]. A top priority was to explore the short- and long-term outcomes of children treated with surgical or non-surgical care for Perthes' Disease. BSCOS carried separately carried out a priority setting exercise amongst surgeons, covering the whole of children's orthopaedics, in which surgical or non-surgical care for Perthes' Disease was the second highest priority [33].

A clinical trial comparing surgical and non-surgical intervention was the best way to address this uncertainty. However, a barrier to conducting such a trial was that non-surgical care was not standardised. Between 2018 and 2020, prior to this doctoral fellowship, the researcher completed a pre-doctoral fellowship in which three key

pieces of work took place to justify the need to optimise the non-surgical treatment of Perthes' Disease. The first was a case review, described briefly in section 1.2.3, which described non-surgical variation of care in the UK [28]. The second piece of work was a systematic review of the non-surgical treatment of Perthes' Disease [34]. This systematic review is discussed in more detail in Chapter two. To summarise, this review demonstrated that there was no robust evidence to support the use of any non-surgical treatment approach compared to another and that further research was needed to optimise non-surgical care. The final piece of work was the beginning of the researcher's engagement with Patient and Public Involvement (PPI) groups. The activities are discussed in more detail in the section below.

1.2.5 Patient and Public Involvement

PPI is widely recognised as beneficial to health research and is a mandatory element of research funded by organisations such as the National Institute for Health and Care Research (NIHR). The benefits of PPI activity in health research have been demonstrated by early career researchers including those completing doctoral level research [35]. Similarly, reflections from researchers and PPI contributors have also demonstrated how PPI engagement can lead to increased impact and have positive influences on research quality [36].

During the application stages for this doctoral fellowship, the researcher visited a NIHR-funded PPI group called the Young Persons Advisory Group (YPAG) [37]. Through this affiliation, children as young as four years old were able to comment on the design of the doctoral fellowship application. It was here that the work from the pre-doctoral fellowship was presented and discussions around current care provision highlighted that physiotherapy intervention needed attention. Children and family members offered insight into their experiences of receiving physiotherapy input for Perthes' Disease as well as other conditions. This first-hand experience from patients and their carers is where the concept of a digital self-management intervention (an app) was born. Individuals recounted experiences of physiotherapy and highlighted

that self-management approaches were not fit for purpose, some called approaches to exercises at home “boring”.

Individuals who took part in early YPAG/PPI activities were invited to be members of a Project Advisory Group (PAG) that met regularly over the course of this doctoral fellowship. The culmination of these PAG and PPI groups are responsible for key milestones/outputs of this project including the acronym and logo for the project. They have also inputted into the design of the intervention and study materials as well as dissemination materials after each study.

The need to deliver this project to develop a digital self-management intervention for the non-surgical treatment of Perthes’ Disease was influenced substantially by the input of PPI/YPAG and PAG members. Their input was invaluable, and in the chapters that follow, the engagement with PPI members and the PAG are described specific to that particular piece of work. In the final chapter, there is a PPI section which provides a clear outline of their meaningful contribution to this project, and their prospective input moving forward.

1.3 Epistemological and ontological position of the researcher

A research paradigm is often considered in a doctoral project, and can be described by considering the views of the researcher in relation to ontology and epistemology. These views can then be used to guide how research takes place, and how the knowledge is conceptualised within relevant communities. Ontology is the study of being and reality, specific to health research, this refers to the nature of reality being studied, i.e. “what can be known?” [38]. Epistemology is the study of knowledge, i.e. “how do we know what we know?” or “how can we study what can be known?” [39]. The two are interlinked, and should align in order to produce coherent and meaningful research [40]. For example in Perthes’ Disease, if one believes that management of the condition is purely biomechanical and can only be managed using physical interventions, then using interviews to study it would be ineffective. Conversely, if one believed it was purely determined by behaviours of children with

Perthes' Disease and their families, and focused only on physical measures such as radiographs or ROM in research, there would be a similar imbalance. An individual's ontological and epistemological position often underpins research paradigms. One of the approaches is pragmatism, which is the approach taken by the researcher throughout the course of this project [38]. Pragmatism is based on the principle of applying the most appropriate methods to explore the problem at hand and is common in studies that apply a range of methods [41, 42].

In the context of this doctoral project, it allowed the researcher to use a range of methods which addressed the specific aims and objectives of each of the studies. In recent research, the roles of patients in research have been described in relation to pragmatism [38]. In a review of the literature surrounding pragmatism in patient-oriented research, Allemang et al produced a table outlining research paradigms and comparing components such as ontology, epistemology, methodology and patient involvement. In Table 1.1 below, this has been adapted to include a column outlining how each of the components were addressed in this doctoral programme of work.

Table 1.1 – Key components of pragmatism in this project*

	Pragmatism	**Evidence in this project**
Ontology	Reality is renegotiated and interpreted based on usefulness in specific contexts	Non-surgical treatment decision making is influenced by the child +/- family input relating to their experiences
Epistemology	Transactional realism: knowledge constructed based on interactions between people and their environments	Key stakeholders (children with Perthes' Disease, their families and clinicians who care for them) are involved at multiple stages of the project
Methodology	Mixed-methods; action-oriented inquiry; design-based	A mix of quantitative and qualitative methods, selected at each individual study to meet the aims of each
Patient roles in research	Consult, involve, collaborate, lead/support (e.g. patients sit on a standing advisory council	Children with Perthes' Disease and their families included as PAG members

	for a clinical trial, or patients involved as research partners)	and involved at every stage of the project

Adapted from Allemang, B., K. Sitter, and G. Dimitropoulos, Pragmatism as a paradigm for patient-oriented research. Health Expect, 2022.* ***25**(1): p. 38-47.*

In each of the subsequent chapters in which a study is presented, there is a reference to the methods and how they align with the ontological and epistemological views of the researcher. Where there is specific consideration of the viewpoint of the researcher and how their stance/views may influence the research, a section called 'Positional reflexivity of the researcher' has been presented. This concept involves providing an insight into how an individual's position and role can impact the current situation [43]. As discussed in Chapters three and six, the researcher presents the challenges of delivering a doctoral project in children's orthopaedics as a specialist physiotherapist. It was important to consider how the researcher's background, such as professional experiences, personal beliefs, and cultural context, shaped the research conducted. For example the researcher's training and experiences in children's orthopaedics have influenced their understanding of the needs of children with Perthes' Disease. The potential is that this experience can lead to biases in interpreting data or feedback. In each study, the steps taken to mitigate this bias are described, ensuring that the findings reflect the genuine experiences of the participants.

The process of developing this digital self-management intervention for children with Perthes' Disease, delivered using a smart device application followed an established approach. The Medical Research Council Framework for the development and evaluation of complex interventions [44]. The framework is described in more detail in Section 2.5.4.1.

1.4 Aims and objectives

Below are the aims and objectives of the project, as well as an exploratory hypothesis that was to be explored.

1.4.1 Aim

The aim of this doctoral programme of work was to utilise the experiences and recommendations of key stakeholders, to deliver a digital self-management intervention for the non-surgical treatment of Perthes' Disease and to assess its usability and acceptability in preparation for a definitive clinical trial.

1.4.2 Objectives

The objectives were to:

1. Investigate the experiences and understanding of children, their families, and clinicians when considering the non-surgical treatment of Perthes' Disease.
2. Develop clinical consensus recommendations for the non-surgical treatment of Perthes' Disease
3. Develop a theoretically grounded, evidence-based, digital self-management intervention (the NON-STOP app) for children with Perthes' Disease and their families
4. Test the acceptability and usability of the NON-STOP app for children with Perthes' Disease and their families, and to clarify any methodological uncertainties prior to the undertaking of a definitive clinical trial.

1.4.3 Exploratory hypothesis

In order to demonstrate a working hypothesis for this project, an exploratory hypothesis was developed. Exploratory hypotheses are commonly used in research areas where there is limited existing knowledge or understanding, such as in the non-surgical treatment of Perthes' Disease [45]. Rather than making definitive predictions, this type of hypothesis seeks to identify patterns, relationships or factors that may influence a phenomenon [46]. In the context of this research, which aimed to develop a digital self-management intervention for the non-surgical treatment of Perthes' Disease, the following was proposed:

Through application of a mix of research methods, it would be possible to develop and preliminarily test a digital self-management intervention for the non-surgical treatment of Perthes' Disease.

1.5 Thesis structure

The thesis is composed of seven chapters, with the content of chapters 2 to 7 described below.

Chapter two: Narrative literature review

In this chapter, a narrative review of the literature regarding surgical and non-surgical treatment of Perthes' Disease is provided and discussed in relation to this project. A review of the literature relating to the development of digital health interventions and the outcomes used in this project is also provided. The chapter concludes with a description of the methodological approaches within this thesis, where an overview of each methodology used in the project is presented.

Chapter three: Exploring the experiences and understanding of key stakeholders

This chapter presents the first of the studies that took part in the doctorate, a qualitative study involving children with Perthes' Disease, their families and the clinicians that care for them. Through qualitative interviews, the researcher presents their experiences and understanding of care regarding the non-surgical treatment of Perthes' Disease. Here the participants also discuss the potential direction for future care and discus positive elements that could be included in a digital self-management intervention for the non-surgical treatment of Perthes' Disease. In reference to the MRC framework, this study maps to the "Developing the intervention" stage of the framework (shown in Figure 2.5).

Chapter four: Clinical consensus recommendations for the non-surgical treatment of Perthes' Disease

In chapter four, the second empirical chapter is presented. A modified Delphi study which resulted in clinical consensus recommendations for the non-surgical treatment of Perthes' Disease. Participants included children's orthopaedic specialists from a range of disciplines including physiotherapists, surgeons and clinical nurse specialists. The participants provided their level of agreement against 87 statements which were presented in an online survey, designed with a survey advisory group made up of experts in the field of children's orthopaedics. The findings from this study devised the bulk of the clinical content for the digital self-management intervention. In reference to the MRC framework, this study maps to the "Developing the intervention" stage of the framework (shown in Figure 2.5). This, after having re-visited the "Core elements" and refined the programme theory.

Chapter five: Creation of the digital self-management intervention: The NON-STOP app

Chapter five presents the process through which the NON-STOP app was created. This presents the intervention development approach used to produce the digital self-management intervention. The impact of psychological theories that underpin behaviour change are also discussed. Here the researcher also presents practical considerations regarding the development of digital health interventions such as contracting, procurement and user testing. Finally, practical examples of how the findings from the project were translated into the NON-STOP app are provided along with images demonstrating the intervention. In reference to the MRC framework, this study maps to the "Developing the intervention" stage of the framework (shown in Figure 2.5). This, after having re-visited the "Core elements" and engaged stakeholders, identified key uncertainties and addressed economic considerations.

Chapter six: Testing the usability and acceptability of the NON-STOP app

Chapter six presents the third and final empirical study, a mixed-methods study to test the usability and acceptability of the NON-STOP app in a small number of children with Perthes' Disease and their families. The study consisted of a before and after observational study, in which the NON-STOP app was given to children from three sites in the UK for six weeks. Outcomes were collected pre- and post-testing using online surveys and consisted of patient-reported outcome measures and measures of usability. Data were also collected relating to their levels of use and engagement directly from the app. In order to provide a more detailed evaluation of the usability and acceptability, a nested focus group study took place. A sub-set of participants who had used the NON-STOP app were invited to take part in focus groups to discuss their experiences of using the NON-STOP app. The discussions that took place were based on the findings of the before and after observational study relating to things like frequency of use and barriers and enablers to using the NON-STOP app. These findings were used to support refinements to the app content to improve its usability and acceptability. In reference to the MRC framework, this study maps to the "Feasibility" stage of the framework (shown in Figure 2.5).

Chapter seven: Discussion

In the final chapter, the project is concluded with a summary of the key findings presented and a revisit of the aims and objectives of the project. The strengths and limitations of the doctoral programme as a whole are considered. The chapter considers the key points related to the challenges and opportunities concerning the further implementation of the NON-STOP app and the impact of the project as a whole on policy and practice. Recommendations for future research are presented by the researcher prior to a final conclusion of the study.

Chapter 2 – Narrative literature review

2.1 Introduction

In this chapter, the literature relevant to the management of Perthes' Disease is reviewed, with a focus on the non-surgical treatment of Perthes' Disease. Following this there is an overview of literature and evidence that was used to provide an understanding of key aspects of developing a digital self-management intervention for affected children. At the end of the chapter, a summary of the available evidence is provided as well as reference to the aims and objectives of the thesis.

2.2 Management of Perthes' Disease

Irrespective of the approach taken, the therapeutic target when managing Perthes' Disease is 'containment'. Containment refers to maintaining the position of the femoral head within the acetabulum, to reduce abnormal stresses to the femoral head which can cause deformation [47]. Containment is crucial in Perthes' Disease, particularly in the early stages of the disease where the femoral epiphysis becomes necrotic and vulnerable to deformation. Maintaining containment allows the femoral head to re-ossify in the later stages of the condition in a position that maximises joint congruency at skeletal maturity.

Approaches for containment can be categorised into two key types, surgical containment and active containment. Surgical containment alters the anatomy of the hip joint to optimise the position of the femoral head within the acetabulum. Surgical approaches are discussed in more detail below. Active containment refers to non-surgical approaches, which aim to keep the child active whilst ensuring that the femoral head remains well situated within the acetabulum. These are also discussed in more detail below.

2.2.1 Surgical management of Perthes' Disease

Surgical containment is most commonly done using a varus osteotomy to the femur [48]. In this approach the surgeon will make a cut to the femur and redirect the femoral head into the acetabulum at an angle to facilitate containment. This surgical approach is shown in Figure 2.1 [49]. The femoral osteotomy is held in place with metalwork until the bone heals. In this surgical method, the metalwork needs to be removed in a later operation. A key advantage of this operation relate to its familiarity in terms of surgical approach. The femoral osteotomy is commonly performed for children with orthopaedic disease such as neurological or neuromuscular conditions, like cerebral palsy, in which hips can sublux or dislocate [50].

Figure 2.1 – Varus osteotomy of the femur shown in radiographs*

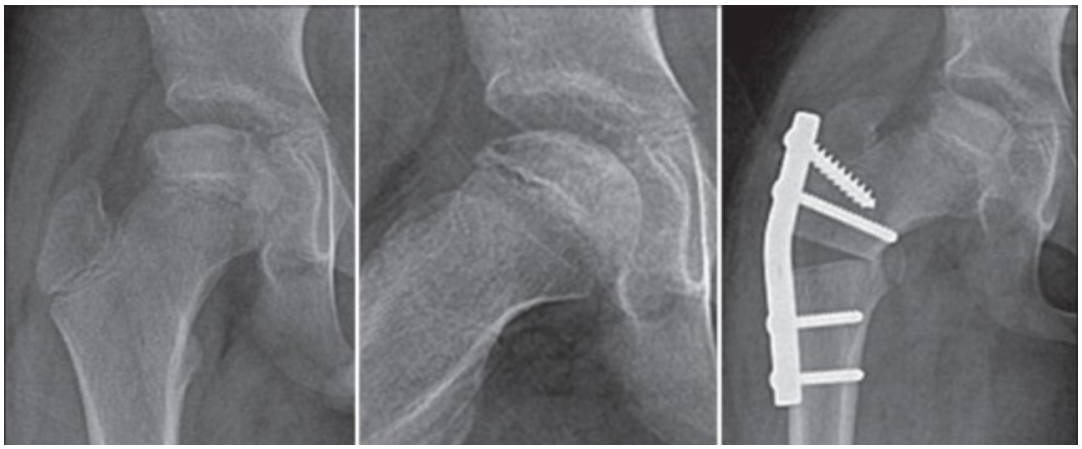


Image from Joseph, B., Management of Perthes' Disease. Indian J Orthop, 2015.* ***49**(1): p. 10-6.*

Changing the shape of the femur does result in some unintended consequences, such as a leg-length discrepancy, though studies with long-term data have demonstrated a shortening of only around 0.5cm at skeletal maturity in the affected limb [51]. Clinically, this level of shortening would not warrant further surgical intervention to correct and this has been discussed extensively in the literature [52]. A review in 2020 compared outcomes of many studies and advised that for less than 1cm difference, no intervention was necessary. Long term data for varus osteotomy in children with Perthes' Disease suggested that in certain groups of patients (older children), the operation improved sphericity of the femoral head [53].

Another commonly used operation, is an acetabular shelf procedure, shown in Figure 2.2 [54]. In this, changes are made to the acetabulum to provide more coverage to the femoral head [55]. It adheres to the same principles as a varus osteotomy, in minimising the stresses to a developing femoral head [56]. In this operation, a bone graft is harvested from the ilium and used to extend the lateral portion of the acetabulum [57]. Increasing the coverage of the acetabulum allows the femoral head to remain within the socket of the joint, optimising joint congruency as the femoral head remodels with reduced extrusion and increased sphericity [58].

Figure 2.2 – Shelf acetabuloplasty shown in radiographs*

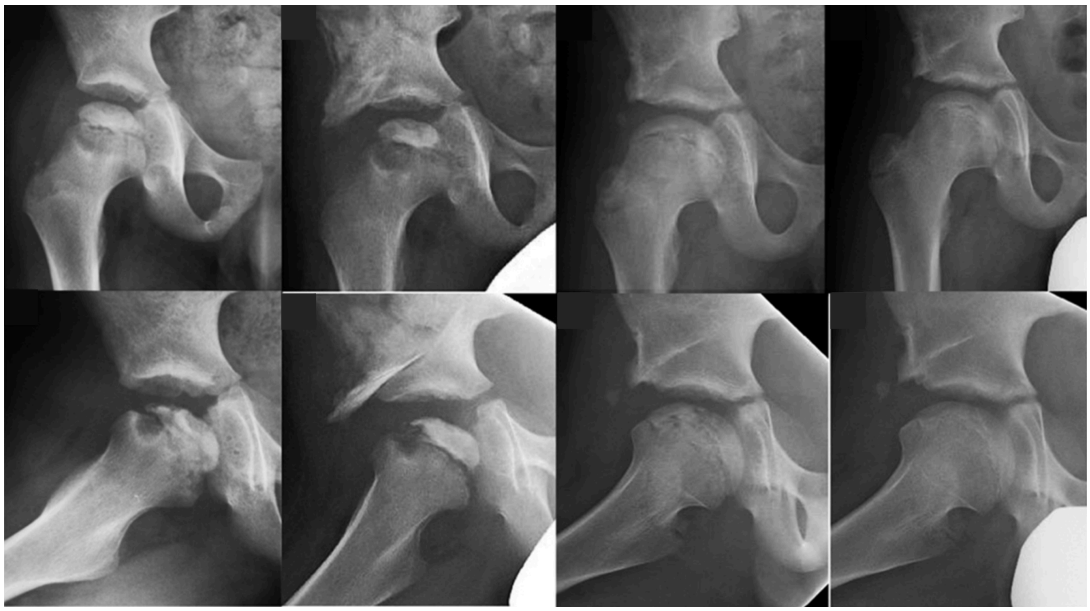


Image from Parmentier, C., et al., Shelf acetabuloplasty in Perthes' Disease: comparison with nonoperative treatment. Current Orthopaedic Practice, 2016.* ***27**(4): p. 375-381.*

Surgical interventions involving bony changes are often combined with soft tissue releases. Soft tissue releases involve cutting the tendons of muscles that may worsen extrusion of the femoral head due to being tight. Whilst often combined with bony surgery, soft-tissue release is sometimes done independent of bony surgery and combined with bracing or cast [59, 60]. The release of tight structures is thought to improve the hip range of motion (ROM) and optimise containment.

2.2.2 Non-surgical treatment of Perthes' Disease

Non-surgical approaches aim to achieve containment of the hip joint without the use of surgery. Many approaches are focused on achieving and/or maintaining acceptable ROM to reach a desired level of containment. Non-surgical approaches include activity modification, bed rest, bracing, casting, traction and physiotherapy exercises. However despite many studies existing exploring the effectiveness of these methods, no clinical guidelines or robust evidence to direct non-surgical treatment exist.

The aims and objectives of this thesis have been described in Chapter one. Whilst these are related to the development and preliminary testing of an intervention for the non-surgical treatment of Perthes' Disease, there was a decision-making process that took place prior to the development of the thesis aims and objectives. This included exploring the available literature surrounding the non-surgical treatment of Perthes' Disease. Prior to the commencement of this doctoral programme of work, the researcher completed a systematic review of the non-surgical treatment of Perthes' Disease [34]. The review compared non-surgical approaches to each other, as well as to surgical interventions and also to no intervention. "No intervention" has been referred to as "active observation" or a "watch and wait" approach [29, 34] and refers to patients who had no formal intervention which essentially allowed the disease to run its natural course.

A summary of these findings is provided below. Non-surgical approaches were categorised into two main groups. The first group investigated orthotic interventions (such as casts, slings and braces), and the second group compared physical interventions (such as strengthening or stretching exercises).

Orthotic management

The orthotic management approach varied. Typically this was an orthotic device to passively maintain abduction of the hip, with the intention to maintain ROM at the hip, and optimise containment. The systematic review included nine studies, which used four different orthotic devices. Some studies were able to demonstrate similar radiological outcomes for children that were managed with either orthotic

management or no/surgical intervention. However most presented no statistically significant differences between the treatment groups in the primary outcomes (radiological). Secondary outcomes included ROM, gait disturbance and quality of life scores. There were no statistically significant differences in these parameters between the group managed with orthotic devices or no/surgical intervention.

Physical intervention

The review also demonstrated inconsistent results when comparing participants treated using physiotherapy interventions (ROM and/or strengthening exercises) or weightbearing modifications in relation to radiological outcomes. These were related to functional outcomes such as ROM and strength rather than radiological outcomes. For example, in the study by Brech et al, children who received a physiotherapy regime of stretching and strengthening exercises had improved levels of ROM and strength compared to those who received no intervention [61].

Summarising the findings of the systematic review

The systematic review was bound by two key factors that influenced the application of the findings. These were poor methodological quality of the included studies, and poor reporting of the interventions used in the included studies.

Each of the included studies were assessed using the Newcastle Ottawa Scale (NOS) [62]. This assessment tool uses three domains: selection, comparability and outcome, to assess the quality and the risk of bias in non-randomised studies. Scores were presented on a scale of zero to eight, with a score of five or less indicating a moderate or high level of bias identified. In this systematic review, more than half had a score of five or less. Most commonly, inadequate follow up and a lack of control for potential confounding was highlighted.

In the review, a similar methodological assessment, the Template for Intervention Description and Replication (TIDieR) checklist was used to present the measure of completeness of intervention reporting [63]. Using this tool, points were awarded to demonstrate that elements of the intervention have been described in the study, i.e., the higher the score, the more complete the reporting. In this systematic review of non-surgical treatment of Perthes' Disease, interventions were generally poorly described. Out of a maximum of 24 points, scores did not exceed 14. A lack of reporting of the intervention materials and intervention dosage was a common concern.

The issues that were observed in the review are not dissimilar to those presented in the small number of studies that have prospectively observed treatment approaches and are considered to be studies of importance in this population [29]. In the study from Herring et al, which compared surgery, with no treatment, brace treatment and physiotherapy, there are issues with quality in terms of potential selection bias due to recruitment taking place at one centre with no randomisation. For example, there are risks that the included participants do not demonstrate the patient population, and that factors such as disease severity or socioeconomic factors could influence results. Similarly with Schoenecker and Rich with the use of A frames (orthoses) and adductor tenotomy [64]. The treatment approaches used demonstrate reasonable clinical findings such as radiographic outcomes, however Patient Reported Outcome Measures (PROMs) are not considered in either study.

2.2.3 Summary of available evidence

The issues presented in the available literature are consistent. There is no robust evidence to suggest that any non-surgical treatment method is superior to another. Given lack of robust evidence on the optimal non-surgical management approach to Perthes' Disease, this thesis sought the experiences and recommendations of key stakeholders, to identify preferences to promote patient self-management involving exercises and education.

2.3 Developing a digital self-management intervention for children

There are no available studies describing the development of a self-management intervention for children with Perthes' Disease. Literature searches to review evidence of similar studies with children with long-term musculoskeletal or orthopaedic conditions also returned nothing. In the absence of evidence relevant to the patient population, literature exploring digital, self-management interventions in the child-health space were examined.

As well as ensuring the methods adhered to the approach set out in this doctoral thesis, it was important to consider how the content of any intervention may be derived. It was also important to consider how any digital intervention may comply or hold relevance to organisations that produce clinical guidelines. An example relevant to this project is discussed below using the National Institute for Health and Care Excellence (NICE) evidence standards framework (ESF) for digital health technologies (DHT).

2.3.1 Self-management

Self-management has been defined as 'the practice of activities that individuals initiate and perform on their own behalf in maintaining life, health and well-being' [65]. In 2003, Lorig et al explored the history and mechanisms of self-management, highlighting its importance in individuals with chronic conditions, such as Perthes' Disease, in managing one's health [66]. The authors discuss behaviours that must be adopted in self-management, which include activity modification, which is similar to this PhD, to develop a self-management intervention for children with Perthes' Disease. The target behaviours for self-management were identified using the studies in this programme of work, such as the first qualitative study, presented in Chapter three. This study produced insights from key stakeholders regarding elements of self-management such as engagement with advice. Further behaviours were identified in a clinical consensus study, presented in Chapter four, these behaviours included activity modification, physical activity adherence (completion of physiotherapy exercises) and education to enhance the individual's understanding of the condition.

Self-management is relatively well accepted in healthcare, with organisations such as NHS England advocating its use in many long term conditions [67]. Successful self-management has the potential to maximise clinical outcomes for patients, whilst reducing treatment costs in the long-term [68]. As well as system-level benefits, it also poses potential benefits for patients with increased levels of independence and control of their condition. There are potential pitfalls, which include nonadherence, and in turn, poorer health outcomes. Striking a balance when designing and developing a self-management intervention requires consideration of a multitude of factors. One in particular, is ensuring that that the population of interest is involved in the process at various stages of design, development and testing.

Often self-management interventions focus on elements of care that can be carried out without medical supervision, but also require routine and frequent completion. It is, therefore, common in disciplines such as physiotherapy, in which self-management i.e., home exercise programmes, are common practice [69]. Self-management has also been effectively demonstrated in respiratory and cardiac conditions [70, 71]. In these conditions, self-management has included independent medication administration. In Perthes' Disease, there are no existing interventions designed for self-management. Typically children are reviewed with relatively long time periods between orthopaedic outpatient appointments, often between three and six months. Providing a self-management intervention has the potential to address concerns and issues from children with Perthes' Disease and their families, allowing them to have a more positive experience by independently managing their condition.

Based on previously completed PPI work by the researcher, a digital self-management intervention for the non-surgical treatment of Perthes' Disease was an appropriate step to take. Self-management in the context of the NON-STOP app is discussed in more detail in Chapter seven.

2.3.2 Digital interventions for physical activity in children

Digital interventions are an increasingly popular method for improving health behaviours, including physical activity in children [72]. In order to develop an effective digital intervention, the content must be specifically designed to meet the needs of the users. In context to this doctoral programme of work, the digital intervention needed to align with the behaviours and motivational influences of both the children with Perthes' Disease and their families, whilst being underpinned by an appropriate theory which is discussed in section 2.5. To do this, relevant literature on the development of digital interventions that impact physical activity in children was reviewed. The review highlighted essential components relating to content of interventions and methods used which promoted the desired outcome, such as increased levels of physical activity. Key elements such as barriers/enablers, fidelity and acceptability of digital interventions were also considered.

An umbrella review by Mannocci et al [73] aimed to summarise the existing literature by reviewing systematic reviews of studies which targeted increased levels of physical activity in children. The review included sub-sections titled "Family and home setting" and "e-Health interventions", which are particularly relevant given the target population (children with Perthes' Disease and their families). Within the "Family and home setting" section, the interventions described in four systematic reviews were not explicitly digital however did include key strategies that could be adapted for digital formats [74-77]. The strategies included goal setting, positive reinforcement and education, all of which align well with the theoretical underpinning of the project, particularly the Self Determination Theory (SDT) which is described in more detail in section 2.5. To summarise, the theory emphasises supporting the autonomy, relatedness and competence of the individual in order to optimise motivation and lead to a sustained change in behaviour [78].

Within the "e-Health interventions" section, there were six systematic reviews which explored digital interventions targeting participants up to 18 years old [79-84]. Whilst the reviews did not include children with Perthes' Disease, there were similarities in the participants from an ability perspective. The reviews included children who were generally mobile, and capable of increasing physical activity however did not meet

the recommended levels of physical activity. The studies involved various types of digital interventions, ranging from applications designed to track or encourage activity [80-84] to web-based or mobile platforms which used reminders and prompts to encourage engagement with exercise routines [79, 80, 82]. Each of the six reviews included step-count as a measure of activity, referencing the use of accelerometers to capture the data. Additionally, three reviews assessed intervention fidelity [81, 82, 84], while four examined acceptability through the use of satisfaction questionnaires or other patient-reported outcomes [81-84]. One review actually referenced the psychological theory used in this doctoral project, SDT [83]. The authors adapted elements of the theory into an outcome measure completed by participants to assess critical motivators such as enjoyment, autonomy, relatedness, and competency.

Several of the reviews within the umbrella review identified barriers and enablers to intervention adherence, which were an important consideration in this doctoral project. For instance, Lau et al discussed barriers related to behavioural factors impacting engagement with the intervention [80]. McIntosh et al highlighted school obligations as a potential barrier to increasing physical activity levels in secondary school students [81]. Finally Monroe et al found that two or fewer notifications per day were perceived as an enabler, improving engagement without overwhelming the user [82]. Once again, these insights were useful in the development of a digital self-management intervention for children with Perthes' Disease and their families. They informed practical decision choices including goal setting, positive reinforcement and reminders to engage with the intervention.

Quality assessment of the systematic reviews included in the umbrella review by Mannocci et al was conducted using Assessing the Methodological Quality of Systematic Reviews (AMSTAR) [85]. AMSTAR is a widely accepted tool for this process [86, 87]. In the umbrella review, Mannocci et al did identify low methodological quality in the majority of the included studies. Whilst there is an argument that this invalidates the findings, there are still contextual benefits to the umbrella review such as using insight from applying approaches from studies that have focused on motivation and positive reinforcement in digital interventions [83]. The low quality also led to the authors highlighting the need for more rigorously designed studies

when evaluating digital interventions, which was helpful in the design of this doctoral project. Particularly given that the findings from this umbrella review were largely to understand the work that had taken place and provide a degree of guidance rather than use as conclusive evidence.

The collective findings suggest that digital health interventions may have potential to be effective in increasing physical activity levels in children. The key strengths are that in the development and review of interventions, theoretical underpinning was present. There were limitations including methodological quality, and also that the included studies did not include any detailed descriptions of user experiences and intervention components. All of which reinforced the need to incorporate this in the development of the NON-STOP app. Overall, the umbrella review confirmed that there are no proven frameworks for the development of digital interventions for children with Perthes' Disease. That being said, there were some meaningful insights to be drawn from the work by Mannocci et al relating to components of the digital self-management intervention for the non-surgical treatment of Perthes' Disease. These included the consideration of usability and acceptability assessment approaches and barriers/enablers to engagement with the intervention in the feasibility study in Chapter six.

2.3.3 NICE Evidence Standards Framework for Digital Health Technologies

In 2019 NICE released the first iteration of the Evidence Standards Framework (ESF) which provided standards to allow people to evaluate Digital Health Technologies (DHT) in relevant settings [88]. Since the outset of this doctoral project in 2021, it has undergone two reviews with subsequent feedback from stakeholders. The latest version of the ESF, updated in 2022, was completed to update the framework in order to include artificial intelligence work [89]. The aim of the framework is to describe the evidence that should be produced in order to demonstrate the value of a DHT has within the UK healthcare system. There is guidance within the framework on how to apply these standards to any given DHT, in this case, a digital self-management intervention for children with Perthes' Disease.

The initial step is the ESF process is to identify the classification that best describes the function of the DHT. For instance, in this project, the DHT includes elements of behaviour change and self-management, which means that the DHT sits in tier 3a [89]. This classification then determines the evidence standards required, which are divided into “minimum evidence standards” and “best practice standards”. These include elements such as “use of appropriate behaviour change techniques” and “demonstrate acceptability with users”. After identifying the evidence for effectiveness standards, contextual questions are asked to identify any risks within the DHT. There are questions about the population of interest, questions about consequences of a DHT that does not perform and any financial or practical burdens of the DHT. In the context of this project, the risks were low. Whilst children are classed as a vulnerable group, which can have inherent risks, mitigations were put in place. The mitigations included regular engagement with PPI and meetings with PAG members during the project to discuss their feelings towards the DHT. This plan was informed by PPI work carried out by the researcher prior to the start of the doctorate.

It is important to note, that any DHT which classifies into a tier, must also meet the criteria of the tiers above. For instance, the DHT in this project, the NON-STOP app, classified as tier 3a, so needed to meet the necessary criteria for tier 3a, but it also met the criteria for tiers 1 and 2. There is an extensive list of evidence categories within tier 1 and tier 2, those most pertinent to this project are, credibility with UK healthcare professionals, relevance to healthcare pathways in the UK health system, production of reliable and consistent evidence and safeguarding. In Chapter five there is a clear explanation of how the NON-STOP app meets the criteria of tiers 1 to 3a and provides evidence of compliance with the ESF. Whilst the ESF is often used to evaluate the DHT, for the purpose of this project it was used to influence the development. That is to say that the guidance helped ensure that the digital intervention met the necessary standards to be described as tier 3a.

2.4 Outcome measures

In order to produce valid, reliable and meaningful results from any clinical research, the outcome measures used were chosen carefully. In child-health particularly, outcomes are complex, especially when considering patient-reported outcome measures (PROMs). In this section the challenges and decisions that the researcher faced when selecting outcome measures are discussed.

2.4.1 Development of a Core Outcome Set

A Core Outcome Set (COS) is an agreed, standardised group of outcomes that are to be reported in any trials in a given research field [90]. They are designed using a set process which involves the inclusion of relevant stakeholders such as clinicians, members of the public and patients to select outcomes of importance to a chosen population [91]. The NIHR have clear guidance for their funding streams that it is best practice to have key stakeholders involved in the development of, or selection of outcomes used in research [92]. This guidance is compounded by the suggestion from the NIHR to utilise COSs where they have been established [93]. One benefit of using a COS is that any trials in a chosen condition or population report findings that can be compared with other trials to produce outputs such as meta-analyses. Another is that trials report outcomes that are thought to be of the upmost importance to the people that are most affected by the condition/topic being researched. These benefits have been highlighted by the trauma and orthopaedic population and are recommended by many when considering research studies, and also research programmes [94].

Prior to the start of this doctoral programme, the researcher, as well as a member of the wider research team (Prof D Perry, Consultant Children's Orthopaedic Surgeon) were involved in the development of a COS for Perthes' Disease [95]. The development group included clinicians from around the world, along with patient and family stakeholders and other key stakeholders; such as charity involvement from the Perthes' Association UK (later dissolved with their work continued by STEPS worldwide). Through robust methods, guided by the Outcome Measures in

Rheumatology (OMERACT) guidelines [96], a review of the literature took place, followed by a consensus (Delphi) study and stakeholder meetings. The result was a list of 14 outcomes that were considered the minimum to be measured in high quality studies involving children with Perthes' Disease [95].

The systematic review identified a large number of outcomes reported in the studies that had taken place in the search period (1990-2017). The lack of consistent outcomes measured means that producing meaningful conclusions from systematic reviews is difficult, and meta analyses are all but impossible. The work be Leo et al demonstrated the need for a set list of outcomes to be measured in future research [95].

Included in the COS are outcomes related to patient-reported outcomes focused on pain, quality of life, sleep, educational participation and mobility. There are also clinical measures, such as radiographic progress, and the financial burden of illness to families and the healthcare system. Considering the results of this COS in the context of this thesis, whilst the projects did not involve a trial, the domains within the COS were important to consider. With that in mind, all domains were considered, and included, particularly in the mixed-methods study in Chapter six where more detail for the included outcome measures can be found.

2.4.2 The Patient-Reported Outcomes Measurement Information System

PROMs are also recommended by organisations like NIHR [93]. Work has been completed by NIHR Biomedical Research Centres to highlight the need, and provide guidance on how to use of PROMs in clinical trials [97]. In 2018 Calvert et al produced guidelines for the inclusion of PROMs, with a specific focus on outcome content. Whilst not specific to child-health research, the conclusions from this work draw upon similar elements to that recommended by the NIHR. PROMs help to ensure that the findings of health research are important to those involved, and can in turn inform patient centred care. They are therefore strongly recommended as part of health and care research.

The Patient-Reported Outcomes Measurement Information System (PROMIS) was developed to create reliable measures of patient-reported health status [98]. The measures report across physical, mental and social wellbeing. Domains and are used in various health conditions including paediatric conditions [99, 100]. PROMIS allows researchers to capture outcomes that are important to both clinicians and patients. A key strength of the measures included within PROMIS, is that they employ item response theory (IRT) [101]. IRT can reduce burden for those completing an outcome measure, while maintaining measurement accuracy. The theory is made up of multiple principles, with each question (item) within the measure evaluated based on the difficulty and how well it measures its intended construct. This is explained in more detail below with an example relevant to this project.

The tool that is of particular relevance for Perthes' Disease is PROMIS Mobility (Appendix A)**.** This outcome measure reports on physical function tasks such as "I could get up from the floor" and "I could ride a bike" and asks for a response on a Likert scale from "with no trouble (5)" to "not able to do (1)". It can be completed by the child, or a proxy version is available for parents of children who are unable to complete the measure.

As outlined above, IRT is an important element of PROMIS tools, and PROMIS Mobility exemplifies this. The measure can be carried out using a Computer Adaptive Test (CAT) which selects or deselects questions based on the response from the user. For example, if when completing the PROMIS Mobility, a user reports that they could "I could run a mile with no trouble", the CAT version of PROMIS Mobility would avoid responses to statements like "I could move my legs". The statistical analysis for PROMIS Mobility is discussed in more detail in Chapter six where it was used as part of the usability and acceptability study of the digital self-management intervention. However, it is important to note that because of the IRT, the scores that are generated from the measure remain accurate due to each item being calibrated against a population mean.

In 2020, the International Perthes' Study Group (IPSG) conducted a multi-centre validity study [102]. The authors concluded that the PROMIS Mobility score has construct validity in measuring the Health-Related Quality of Life (HRQoL) of children

in varying stages of Perthes' Disease. In 2021, the PROMIS Mobility score was used to assess the hip function of adolescents previously affected by childhood hip conditions [103]. The majority of the included participants (232/266, 87.2%) had Perthes' Disease. Luo et al demonstrated construct validity of PROMIS Mobility, with a strong correlation between PROMIS Mobility and other PROMs.

In conclusion, PROMIS Mobility provides a robust and patient-centred approach to assessing physical function in Perthes' Disease and has been validated in the patient population. Along with its alignment with the COS described earlier, this makes PROMIS Mobility well suited for inclusion in this doctoral project.

2.5 Methodology

This doctoral programme of work employed a mix of methods over its course. This method aligns well with the pragmatic approach that has been taken during this PhD. The following section outlines the methodological approaches taken at each of the various stages of the project. There is study-specific detail in each of the subsequent chapters, however this section provides an overview of the methods used and justification. This starts with a section describing the psychological theory that underpins the project.

2.5.1 Theoretical underpinning of thesis

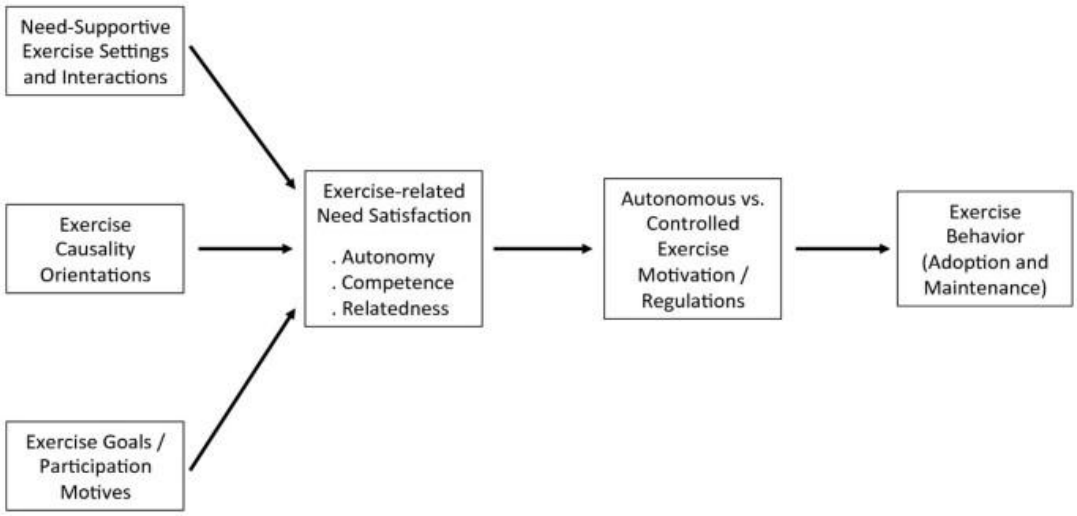
The methods employed throughout the duration of this project were based on two key theories, the Self Determination Theory (SDT) and Socio-Ecological Model (SEM). Both theories consider motivation and the factors affecting the behaviours and actions of people.

2.5.1.1 Self Determination Theory

Given that the overall project aims are to develop a digital intervention to promote engagement in exercise and physical activity for these children, this study draws upon SDT, shown in Figure 2.3 [78]. This psychological theory 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 [104].

Figure 2.3 – Diagram illustrating the key concepts of Self Determination Theory*



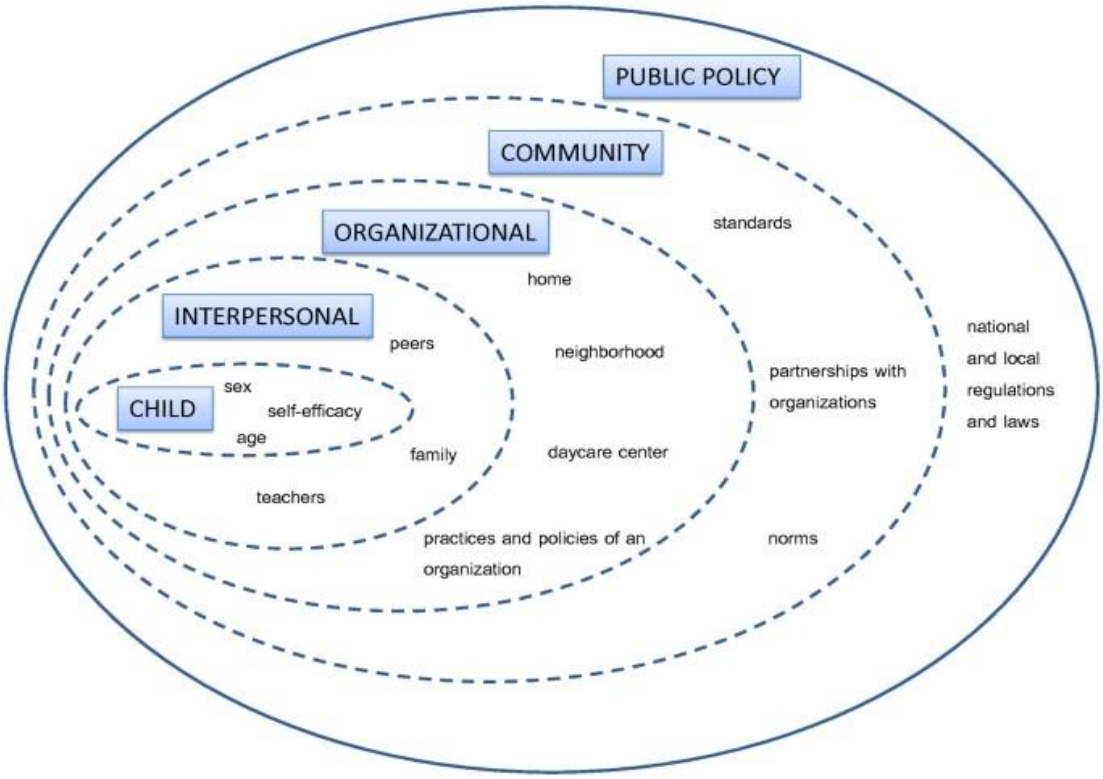
Reproduced from Teixeira, P.J., et al., Exercise, physical activity, and self-determination theory: A systematic review. International Journal of Behavioral Nutrition and Physical Activity, 2012. **9(1): p. 78.*

SDT has been used to support the design of health promotion interventions targeting physical activity and exercise in children, and such interventions have been shown to be effective in the improvement of physical activity outcomes. SDT proposes that focus on the importance of intrinsic motivation is beneficial for exercise and physical activity [105]. Previous literature demonstrates success in terms of increased levels of exercise and physical activity and exercise when the 'psychological needs' have been addressed [106, 107]. Autonomy has been demonstrated in studies in which children and adolescents have designed their own physical activity regime, not dissimilar to approaches used in self-management. This was found to increase levels of autonomy and enjoyment [108]. 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.

2.5.1.2 Socio-Ecological Model

Whilst SDT is the primary theory underpinning this study, it will be further informed in part by the Socio-Ecological Model (SEM) of behaviour [109]. Although SDT seeks to explain the factors motivating behaviour 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. Although five levels of intervention are identified in SEM (see Figure 2.4), in this study there was a focus on three levels: the child (applying SDT); 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.

Figure 2.4 – The Socio-Ecological Model of behaviour*



Reproduced from Mehtälä, M.A.K., et al., A socio-ecological approach to physical activity interventions in childcare: a systematic review. International Journal of Behavioral Nutrition and Physical Activity, 2014.* ***11**(1): p. 22.*

The benefits of an integrated theoretical approach to intervention design, combining SDT with SEM, have previously been demonstrated for an intervention targeting physical activity promotion in adolescents [108]. Indeed, it is widely recognised now that theory integration is encouraged, as it can reduce the redundancy when applying two or more relevant theories and utilise the strengths of specific theories. Zhang and Solmon suggest that the integration of SDT and SEM can provide a unique insight into ways of structuring a supportive environment and build comprehensive interventions to increase physical activity engagement [110].

2.5.2 Chapter three – Exploring the understanding of key stakeholders

In Chapter three, semi-structured interviews took place with key stakeholders to gain an understanding of their experiences of care, and gain insight into what future care, including self-management, should include. The key stakeholders included children with Perthes' Disease, their families and clinicians who provide care for them. Interviews took part with children/family members as pairs, referred to as dyads, and took part with clinician participants as one to one interviews. The data collected as part of the interviews in this study were analysed using the Framework Method [111].

2.5.2.1 Interviews with children

Gaining information from children is something not commonly done in qualitative health research. There are specific challenges that include a variation in language skills and communication abilities that change depending on the age of the child. Other challenges include engagement and rapport building with the child. All of these come with inherent risks methodologically. It is important to ensure that the setting and methods are optimised to make the child feel comfortable and willing to engage [112].

It is possible to suggest that other methods could have been used, both in terms of data collection and analysis. For instance, in data collection, focus groups would have minimised data collection time by bringing participants all together at once, which has time and cost benefits to the researcher [113]. A focus group approach also has the potential for participants to generate ideas and explore their feelings in a self-motivating way as they share lived experiences with each other. Whilst focus groups do pose logistical benefits for the researcher, it is important to understand the associated limitations. Focus groups rely on participants being in the same place, at the same time, responding to the same question(s). Similar to the logistical benefits, there are also logistical difficulties with arranging the focus groups, particularly when involving children from a range of geographical locations. More commonly associated with focus groups, is the risk that participants feel uncomfortable sharing and engaging fully in the group discussions. This is particularly common when discussion

involves sensitive topics, common in health research [114]. In the context of this study, it is reasonable to suggest that feeling uncomfortable was a real possibility when asking children to discuss their experiences of care in front of other children and families. It is also reasonable to suggest that other methods of data collection within interview-type settings could have been used with children.

Previous qualitative research used in child-health research has employed methods such as having children draw pictures relating to their answers and photo elicitation [115, 116]. These methods allow interviewers to build rapport well with children by asking them to express their feelings through play and creative activities. It can make them feel more involved and build rapport. However, these methods have limitations including, but not limited to time and additional resources [117]. They also require the expertise of researchers conducting the interviews, which was not realistic in the scope of the interviewer's experience level at the time of data collection [118].

2.5.2.2 The Framework Method

Framework Method, sometimes referred to as the 'Framework Approach' was developed in the 1980s and was originally used as a way to analyse data from applied policy research. Its development was based on commissioned briefs within social research [119]. It provides a targeted way to analyse qualitative data using a structure in which data can collected, then analysed through categorisation into codes that relate to the study aims and objectives. This can then allow the researchers to translate the data collected into a conceptual explanation of what is being explored [120]. There are discrepancies within the literature regarding the number of analytic stages that exist in the Framework approach [111] however, they have been simplified into five data analysis processes since their initial development [121].

1. Familiarisation – in the first stage of the analysis the researcher familiarises themselves with the data and begins identifying topics of interest, that are typically recurrent across the data, and then organising these to create code names.

2. Coding – the next stage of the analysis organises the data. After becoming familiar with the data and identifying themes, the theoretical framework is constructed by applying code names. It is worth noting that the Framework approach still allows for emergent themes.
3. Indexing – the sections of the data are then labelled according to their themes. This can be done using numbers or words, or even phrases to best identify the data.
4. Charting – this stage involves rearranging the data to create order. This typically involves the creation of a table using the code names to display the theoretical framework to be applied to the data. This often involves summaries that explain the theme and can include excerpts.
5. Mapping and interpretation – in the final stage of the analysis, the table is used to investigate the relationship between the themes and the data. This is regularly done using data management software to categorise the data into codes/themes for easier analysis. At this point, the analysis can be interpreted effectively.

The Framework Method suited the semi-structured interview study because it is designed to structure data collection and analyse data sets that have similar topics or key issues that participants will offer an opinion on or describe [111]. For instance in this study, the topic guide was pre-populated reflecting PPI activity and theoretical basis from previous work. This preparatory activity pre-specified a structure on the data. The participants were asked questions that aimed to have them describe their experiences of clinical care as clinicians delivering care, or as patients/families receiving care. It is possible to suggest that a limitation to the Framework Method is that the coding framework can be limited or constrained by the theoretical underpinning. For example if the theory does not cover aspects of the data that may emerge, it can be difficult to facilitate codes that are not included within the theory. That being said, within this study, the theories used were selected based on previous work in the subject area. The method also suits the pragmatic approach taken within the PhD, but more specifically within this qualitative study. The Framework Method imposes a structure, but also allows new themes to emerge through the analytic

approach. The approach balances a deductive approach, in which the individual explores the elements of the framework, with an inductive analysis, which includes new emerging themes from the data [122]. It has been described as 'not aligned with a particular epistemological, philosophical or theoretical approach' and rather a flexible tool for qualitative studies that generate themes, which this study does.

There are other data analysis methods that could have been employed in this study. Two commonly used are content and thematic analysis. Content analysis examines data and identifies the presence of certain words, subjects or concepts. It can be effective if the data collection was carried out in an attempt to assess answers that are likely to include a certain topics/phrases, for example, as recorded in free-text responses in surveys or questionnaires [123]. This method can lead to a reductive approach to managing the data, in only looking for the presence of concepts/certain topics [124]. This was not the aim of this study, which was exploratory in nature, which was an element of the rationale for opting for the Framework Method. Unlike content analysis, thematic analysis does allow the analyst to infer and interpret the data in more detail [125, 126]. It is possible consider the thoughts and feelings of the participants with more depth. It does so by focussing on the experience of the participant. In the context of this study, this was not necessary given the utilisation of previous work, PPI input and the developed topic guides providing a clear aim and set of objectives to inform the development of a digital intervention.

The Framework Method finds itself somewhere between these two approaches in that it has coding that are informed by literature and theory. This directs the interview questions, yet the analysis is still inductive. Having coding that is amenable to change allows for a more realistic data collection and analysis from the participants [111].

2.5.3 Chapter four – Clinical consensus on non-surgical treatment of Perthes' Disease

A modified Delphi study was conducted in Chapter four and used to produce clinical consensus recommendations for the non-surgical treatment of Perthes' Disease. Delphi studies are one method of ascertaining consensus. There are, however,

alternative methods that can be employed. The most common are Consensus Development Conferences, the Nominal Group Technique (NGT) and RAND University of California Los Angeles (UCLA) appropriateness method (RAM).

Consensus Development Conferences are one-off, face-to-face sessions where a panel of experts are presented with the available evidence on a topic [127]. The evidence is then reviewed separately, and consensus statements are produced by the expert panel. Benefits of this method include efficiency in terms of getting consensus statements quickly after posing them to the expert panel. The downside to this method is that there is often a lack of definition of what constitutes consensus which could impact the validity of the results. Similarly, an open-panel can often have social implications where less-forthcoming individuals may not share opinions, and vice versa which introduces a risk of bias. There are also significant cost implications in terms of travel and organisation. The consistency of the steps within this method is an issue affecting the validity of the results. It has been recommended that formalising the process is necessary to improve the validity of results [128].

NGT involves experts meeting with an independently generated list of statements/ideas. The list is then combined with the other experts on the panel by a moderator and rounds of ranking and prioritisation take place until a pre-determined cut off is achieved [129]. The benefits of this include anonymity which reduces bias and, in contrast to Consensus Development Conferences, allows less-forthcoming individuals to contribute confidently and anonymously [127]. It has been used in a similar patient population [130]. That being said, it is also expensive and complex to organise due to the face-to-face format.

The most similar to Delphi methods is RAM, which is said to combine elements of NGT and Delphi methodology [131]. This method is particularly useful for topics that have existing evidence base due to the requirement of evidence briefing and review which informs the survey/scenarios presented to panel members [132]. The survey/scenarios are then reviewed independently, minimising the risk of bias and given an ‘appropriateness’ rating. After this, a meeting of the panel takes place to discuss group scores. Then finally, a final anonymous round of rating for ‘appropriateness’ with median scores used to decide the appropriateness of a certain

statement. Previously it was necessary to meet face-to-face for RAM methods, however this has changed with technological advances allowing online meetings to facilitate the discussion of group scores [133]. The major limitation for RAM is the challenging language used, particularly in context to this study in which labelling a treatment approach as 'inappropriate' has connotations of unsafety in the clinical setting. The Delphi method allows more flexibility in this domain. A summary of the alternative consensus methods available are seen in Table 2.1.

Table 2.1 – Summary of consensus methods

Method	**Sample**	**Remote delivery**	**Evidence presented**	**Major strength**	**Major limitation**
Delphi [134-137]	6-3000+	Yes	Optional	Possibility of large sample	Long time to complete compared with other methods
Consensus development conference [127, 128]	5-10	No	Yes	Promotes discussion and debate	Risk of bias with under-represented panellists
Nominal Group Technique [129]	5-9	No	Optional	Equal opportunity for panellists to present	Expensive to arrange face-to-face
RAND UCLA Appropriateness Method [131-133]	7-15	Yes	Yes	Mandatory use of evidence synthesis	Challenging language in method

The methodology also offers a certain degree of flexibility for the researchers using it compared to other methods such as the RAND UCLA method in which the methodology is very strict. The term 'modified' Delphi study has become more common in consensus studies, particularly when aiming for clinical consensus on management of a condition [134, 135, 137]. There is, however, very little in the way of describing what constitutes a 'modified' Delphi study, one common theme in a 'modified Delphi' is that there is a steering group for the design of the survey and a

pre-determined number of rounds [136]. This study was a modified Delphi study and employed similar methods for survey design, which is discussed in Chapter four.

2.5.4 Chapter five – Producing the digital self-management: The NON-STOP app

In Chapter five, the development process that took place when creating the NON-STOP app is designed. The methods here relate to the intervention development approach used. Further study-specific detail is presented in Chapter five.

2.5.4.1 Approaches to intervention development

Previous methodological research has summarised the various approaches available in intervention development [138]. In 2019 O'Cathain et al published a systematic methods overview (SMO) of the methods used to develop complex interventions in applied health care settings [138]. The authors, following a robust guidance for undertaking SMOs [139], presented a taxonomy of approaches to intervention development, divided into eight categories (Table 2.2). This taxonomy allows users to understand the various stages of intervention development as well as evaluation and implementation.

For the development of the NON-STOP app, a combined approach of the Medical Research Council (MRC) framework for developing and evaluating complex interventions (hereafter referred to as the 'MRC Framework') and the Behaviour Change Wheel (BCW) was used. A combined approach is common in applied health settings [138, 140]. The overlap of approaches is something that O'Cathain et al discuss as a limitation to the taxonomy they have produced, stating that many of the approaches are similar. The choice to combine approaches aligns well with the pragmatic approach of the researcher shown in Chapter one.

Choosing a single approach to intervention design for children with Perthes' Disease that explicitly incorporates their views and experiences was deemed necessary, but not sufficient. For example, the approach allowed the researcher to incorporate the

views of the children with Perthes' Disease and their families. Combining with the MRC framework and BCW provided comprehensive theoretical and evidence-based rigor; both approaches are complimentary, incorporating user input in the development process, but also place equal value on using the best available evidence and theory to optimise this process. The MRC Framework maps out the stages of intervention development and evaluation, including reference to theory and evidence. The BCW provides additional theory to enhance understanding of intervention components which cannot be found in the MRC Framework. A combined approach in this instance ensured that the NON-STOP was grounded in the best available theory and evidence whilst still considering the views of the key stakeholders.

It would have been possible to select another intervention development approach from the many different options described in the taxonomy, but it is beyond the scope of this thesis to critically appraise each one [138]. Each has their own strengths and limitations. However, two alternative approaches were considered more fully but not adopted in the development of the NON-STOP app. 'Partnership' approaches (Category 1), which involve intervention-users throughout the development process, share a central feature that users have at least equal decision-making powers to the research team. Co-production has proven effective in the design and implementation of school-based public health interventions [141, 142]. The limitations around this method relate to difficulties in ensuring evidence-based content. There are risks of co-produced materials lacking clinical credibility, or even the potential to deviate from clinical practice. With regard to the NON-STOP app, whilst app users needed to be actively involved in the app design through PPI engagement activities, it must be acknowledged that there were important clinical considerations related to app content (i.e. clinical consensus study findings) that might over-ride patient preferences (e.g. if relating to patient safety or availability of clinical support). Similarly, there were other theory and evidence-based approaches (Category 3) that could have been selected but were not. One example considered was intervention mapping, an approach effectively used in public health contexts [143]. Intervention mapping describes six detailed stages from the design and development of the

intervention through to its implementation. Whilst shown to be effective (e.g. the development of self-management interventions relating to physical activity [144]), intervention mapping is relatively resource-intensive, and at times each stage can be somewhat prescriptive [138]. It might also be argued that the MRC Framework includes many of the steps described within intervention mapping, but affords greater flexibility in their application.

Table 2.2 – Taxonomy of approaches to intervention development*

Category	**INDEX team definition**	**Defined approach**
1. Partnership	The people for whom the intervention aims to help are involved in decision-making about the intervention throughout the development process, having at least equal decision-making powers with members of the research team	Co-production, co-creation, co-design, co-operative design
		User-driven
		Experience-based co-design (EBCD) and accelerated EBCD
2. Target population-centred	Interventions are based on the views and actions of the people who will use the intervention	Person-based
		User-centred
		Human-centred design
3. Theory and evidence-based	Interventions are based on combining published research evidence and formal theories (e.g. psychological or organisational theories) or theories specific to the intervention	MRC Framework for developing and evaluating complex interventions
		Behaviour-change wheel (BCW)
		Intervention mapping (IM)
		Matrix Assisting Practitioner's Intervention Planning Tool (MAP-IT)
		Normalisation process theory (NPT)
		Theoretical domains framework (TDF)

4. Implementation-based	Interventions are developed with attention to ensuring the intervention will be used in the real world if effective	Reach, Effectiveness, Adoption, Implementation, Maintenance (RE-AIM)
5. Efficiency based	Components of an intervention are tested using experimental designs to determine active components and make interventions more efficient	Multiphase optimization strategy (MOST)
		Multi-level and fractional factorial experiments
		Micro-randomisation trials
6. Stepped or phased based	Interventions are developed through emphasis on a systematic overview of processes involved in intervention development	Six essential Steps for Quality Intervention Development (6SQUID)
		Five actions model
		Obesity-Related Behavioural Intervention Trials (ORBIT)
7. Intervention-specific	An intervention development approach is constructed for a specific type of intervention	Digital (e.g. Integrate, Design, Assess and Share (IDEAS))
		Patient decision support or aids
		Group interventions
8. Combination	Existing approaches to intervention development are combined	Participatory Action Research based on theories of Behaviour Change and Persuasive Technology (PAR-BCP)

Table adapted from O'Cathain, A., et al., Guidance on how to develop complex interventions to improve health and healthcare. BMJ Open, 2019. **9(8): p. e029954.*

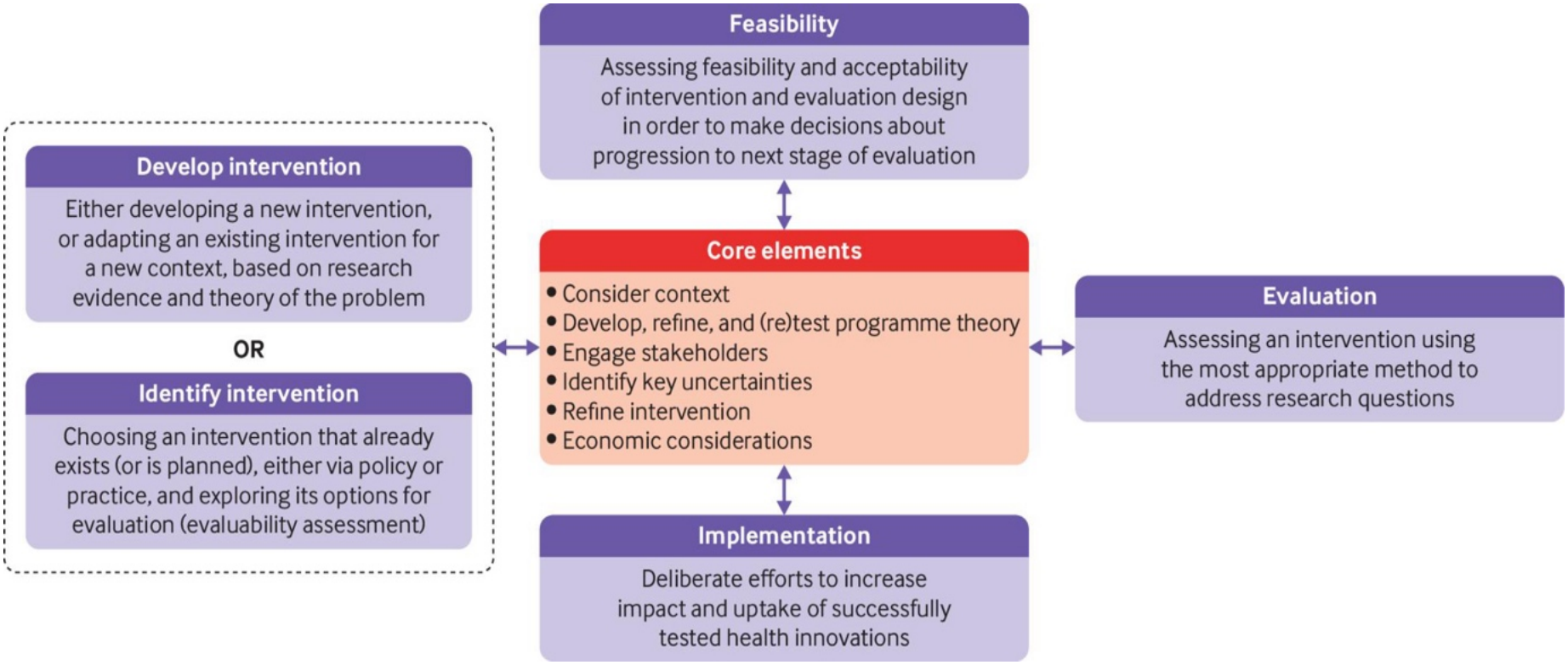
2.5.4.2 MRC Framework

The MRC produced an updated framework for developing and evaluating complex interventions in 2021 [44], broadly describing the development of an intervention in four phases (shown in more detail in Figure 2.5):

- Intervention development (or identification)
- Feasibility testing
- Evaluation
- Implementation

A focus of the MRC guidance is to consider the dynamic relationship of the intervention and its context. To do this, the guidance encourages the user to consider six core elements at each phase. At this point the research team can decide whether to proceed to the next phase, return to or repeat a phase, or stop the project. This iterative approach allows regular review of the core elements at each stage, as seen in Figure 2.5 below. These core elements are reviewed in more detail with specific examples relating to the NON-STOP app in Table 5.1.

Figure 2.5 – The MRC Framework for developing and evaluating complex interventions*



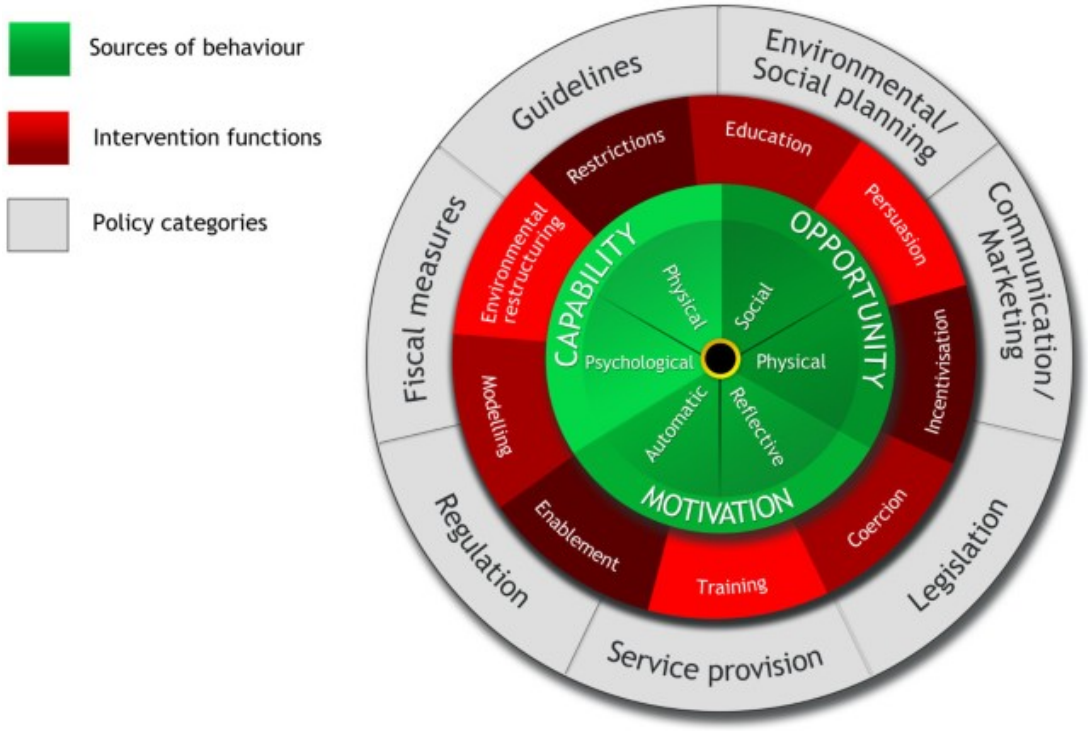
Reproduced from Skivington, K., et al. (2021). "A new framework for developing and evaluating complex interventions: update of Medical Research Council guidance." BMJ **374: n2061.*

2.5.4.3 Behaviour Change Wheel

First described in 2011 in its current format of a 'Behaviour Change Wheel', Michie et al proposed a system for consolidating the existing behaviour change approaches in to a useful framework [145]. The purpose of developing this 'wheel' was to help researchers match behavioural interventions, e.g. physical activity promotion, with their target population. Similar to the MRC Framework it also allows the user to assess the context in which the intervention will be delivered.

The BCW can be seen in Figure 2.6 and is made up of three layers. The innermost layer is made up of the Capability, Opportunity, Motivation and Behaviour model (COM-B), this is discussed in more detail in Figure 5.1. This model is a 'behaviour system' which suggests that for a behaviour to take place, the individual must have the required level of capability, motivation and opportunity. The middle layer of the wheel relates to ways in which the intervention may influence an individual, whether it be positively, i.e. towards performing the behaviour, or negatively, i.e., against performing the intended behaviour. The outer layer comprises the policy categories. These categories relate to implementation, and describe ways in which the chosen intervention may be implemented. For example, through guidelines supported by well-established clinical infrastructures like NICE.

Figure 2.6 – The Behaviour Change Wheel (BCW)*



Reproduced from Michie, S., M.M. van Stralen, and R. West, The behaviour change wheel: A new method for characterising and designing behaviour change interventions. Implementation Science, 2011. **6(1): p. 42.*

2.5.5 Chapter six – Testing the usability and acceptability of the NON-STOP app

Chapter six presents the final study of the doctoral project. A mixed-methods feasibility study to test the usability and acceptability of the NON-STOP app. In the first component of the study, a before and after observational study took place in which children with Perthes' Disease and their families tested the app for six-weeks. In the second component, focus groups were completed to provide detail and insight into the experiences of using the NON-STOP app. Unlike the first study in Chapter three, where personal experiences of Perthes' Disease were described requiring interviews, discussing the NON-STOP app was considered much less sensitive. All users had used the app and to discuss this was more suited to a group discussion, interactions with each other were desired in order to explore a deeper understanding of potential refinements.

2.5.5.1 Mixed-methods

Mixed-methods as a methodological approach are increasingly common in healthcare research, and are used to address a research aim using an integrated method [146]. A common example of this includes combining quantitative elements that demonstrate effectiveness, or 'whether something works' with qualitative elements, that offer nuances as to 'why something works'.

For this final study of this doctoral programme, a mixed-methods study was chosen because it would meet the intended aims of this sub-study. There are a number of approaches within mixed-methods one must consider when conducting this type of research. An 'explanatory sequential' design was deemed to be the most fitting way to describe integration of data collection and analysis in this study [147]. In this design, a quantitative element is followed by a qualitative element. Ivankova et al suggest that this method provides a better understanding of the research problem [147], with the qualitative element informed by the quantitative findings, with the intention of explaining the quantitative findings. In the context of the NON-STOP app, and this study, the before and after observational study enabled use of the app and collection of necessary data to understand the use of the intervention. The nested focus group gave participants who had used the NON-STOP app the opportunity to explain how usable the app was, and why they felt that way. It also allowed them to explain any reasons that hindered intervention-use and suggest changes to the intervention. Using mixed-methods can be considered time-consuming and requires careful planning, but offers a more comprehensive understanding of the intervention. Particularly in this project, in preparation for further testing in a clinical trial.

Similar methods have been used in children's orthopaedic research recently. A randomised clinical trial compared two different interventions for displaced distal radius fractures [148]. The study team set out to provide clinical findings relating to the treatment of the fracture and patient outcomes. These results were quantitative in nature, and recovery was measured using patient-reported outcome measures (PROMs) focusing on upper-limb function and quality of life. In 2024, Phelps et al described the findings from a nested qualitative study which took place as part of the randomised clinical trial [149]. In the qualitative component, parents of children who

took part in the clinical trial were interviewed to provide an overview of their experience of the intervention they received. The authors also explored the perception of feeling “recovered” which provides another example of how mixed methodology can provide a stronger understanding of a phenomenon. The trial clinical findings provide valuable information on treatment effects; however the qualitative component gave a more rounded perspective on care. For instance, parents reported that recovery involved insight into things that mattered most to the child/family such as returning to sport, and improved appearance of the fractured arm [149].

There are alternative approaches that could have been used in this study within the spectrum of mixed methodology designs. One includes the convergent design, in which the quantitative and qualitative data collection and analysis occur simultaneously [150]. For instance in this NON-STOP app usability and acceptability study, it would have required data collection to have taken place during the app-testing period. The qualitative data collection would have needed to be adapted into something like a free-text response via the app to collect information relating to the use of the NON-STOP app. This would have increased the burden to the participant, and could have affected their adherence to using the intervention. Whereas, in the focus groups conducted by the researcher, not all participants are included, minimising burden. Also collection of PROMs collection after the qualitative element may affect findings. The main reason being that participants views may change as a result of reflecting on their experience, rather than any true change in outcome as a result of the intervention itself. Using a convergent method would have had a significant limitation for this project because a single time-point of data collection would not have allowed the quantitative element to inform the qualitative [151]. For instance, the findings could not iteratively inform changes/adaptations to the questions asked in the nested focus group.

2.5.5.2 Before and after observational study

The before and after observational study was chosen for the quantitative component because of the high levels of efficiency it provides when trying to assess criteria relating to an intervention [44]. This design is very common in early phase feasibility work and is often opted for because of its ease of application. It does not require randomisation or a control group which can be time inefficient. However there are some limitations with this methodology. These include the lack of control group to account for things such as confounding and selection bias and is not suitable for estimating intervention effectiveness. Steps to reduce the risk of selection bias were managed using a specific sampling method, as described in the sampling section, seen in Chapter six. The supervisory team for the project of work included intervention development specialists (SR and DK) who shared their expertise when the methodology was discussed and chosen. A suitable alternative to this method would have been to carry out a randomised feasibility trial to compare the use of the NON-STOP app with 'usual care' [152]. Delivering this would have been much more time consuming, require larger sample sizes and was outside of the scope of this doctoral programme of work.

2.5.5.3 Focus groups

The qualitative interview study in Chapter three allowed the researcher to understand participants' experiences of care. Trying to gain an insight of an individual's experiences over the course of a disease process is sensitive. It is important to achieve this without the influence of others and to allow participants to feel safe to share their own personal experiences. Interviews were therefore an effective way of doing this [153]. However, in this mixed-methods study, the aim was to understand something less sensitive, and a focus group offers many advantages by including more participants.

The aim of the app-testing study was to understand the acceptability and usability of the intervention. Creating a space for participants to feel comfortable and allowing them to converse with people who are similar to them (i.e. children with Perthes'

Disease and their family members) allows for more open discussion [113]. It also allows for discussions to include things not planned, which in turn leads to more meaningful data collection. Focus groups allowed for open discussions with others to share their experiences of using the NON-STOP app and consider aspects that they liked/disliked. Users were able to create discussions and provide insights that may not have been covered by the researcher's topic guide. It also allowed participants to discuss aspects of the NON-STOP app that they would alter or adapt, and to discuss how to optimise the intervention based on their recent experiences.

Kennedy et al in 2001 provided guidance on when to conduct focus groups with young children (age 6-12 years) which is relevant to this patient population [154]. Specifically, focus groups are effective when looking to evaluate an intervention and provide rich and meaningful responses from children, which may not always be possible when using 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 were adopted here. These included increasing the comfort of children by ensuring there was a peer audience and to adopt language that is appropriate and applicable to children. Something that, from a reflexive perspective, is very applicable given the researcher's experience as a children's physiotherapist.

It would have been possible to consider interviews for this study, as has been proven in the published methods from chapter three in this project [155]. However, as well as the advantages of the focus groups described above, it would also have been more burdensome for the researcher to carry out individual interviews. This, combined with the potential for less meaningful data, led to the decision to conduct focus groups. Similarly, the choice of focus groups allowed the researcher to develop skills in a data collection method that was new to them. It was carried out with support from the supervisory team who have experience in this field.

2.6 Conclusion

The topics covered in this chapter further support the need to deliver a digital self-management intervention for the non-surgical treatment of Perthes' Disease. The aims and objectives outlined were designed with this in mind, and through the use of appropriately selected methodology, have been addressed in the subsequent chapters.

Chapter 3 – Exploring the understanding of key stakeholders

3.1 Introduction

This chapter reports the first empirical study as part of the doctoral programme of work, a qualitative interview study which explored the experiences and understanding of key stakeholders relating to the non-surgical treatment of Perthes' Disease. The findings have been published as:

> Galloway, A. M., Pini, S., Holton, C., Perry, D. C., Redmond, A., Siddle, H. J., & Richards, S. (2023). "Waiting for the best day of your life". A qualitative interview study of patients' and clinicians' experiences of Perthes' Disease. *Bone & Joint Open*, *4*(10), 735-741. https://doi.org/10.1302/2633-1462.410.BJO-2023-0108.R1

In reference to the MRC framework, this study maps to the "Developing the intervention" stage of the framework (shown in Figure 2.5). The Core elements of relevance at this stage were engaging stakeholders, included as participants and aiming to identify key uncertainties.

3.1.1 Background

Understanding the factors driving variation in the provision of non-surgical treatment and the potential unmet needs is vital. To do this, the experiences and views of children, their families, and clinicians regarding non-surgical treatment of Perthes' Disease in the NHS must be documented. To date, there has been no in-depth qualitative work conducted with key stakeholders experiencing/providing care for children with Perthes' Disease. This study is the first of the researcher's NIHR-funded programme of work intended to address this important gap. In the wider context of this PhD, this study informed the overall aim of the project which is to utilise the experiences and recommendations of key stakeholders in the development of a digital self-management intervention for non-surgical treatment of Perthes' Disease. There are more detailed aims and objectives for this study described below.

In addition, this study discovered stakeholder views around the potential for a new digital intervention for Perthes' Disease aimed at improving a child/family's ability to self-manage the condition. An important part of this self-management will be promotion of exercise and physical activity. This has many benefits, amongst which are improvements in the strength and stability of children affected by Perthes' Disease [156]. The design of this qualitative study allowed the researcher to identify potential intervention functions that meet the needs of the patient population, i.e. development of a digital self-management intervention for children with Perthes' Disease. There are also potentially wider health benefits such as reducing the burden of childhood obesity [157]. This is particularly relevant 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 [158]. This study engaged with a wide range of people, both healthcare-professional and otherwise, to understand to what extent children/families are currently involved in the decision-making process concerning their care and how a digital intervention might enhance this important aspect of self-management.

3.2 Previous qualitative research in Perthes' Disease

Searches were carried out in an attempt to find previous studies that had directly explored the views of children with Perthes' Disease, their families, or the clinicians caring for them. The focus was on non-surgical care and topics considering future interventions. No studies were found.

Further searches were completed to identify relevant literature. They were completed at the time of writing the study protocol (October 2021), using recommended databases including PubMed and MEDLINE. Terms included "Perthes' Disease", "experiences", "interview" and "qualitative". Searches identified studies that were related in terms of patient population (children's orthopaedics), or similar

methodologies (interviews with key stakeholders) were identified and are discussed below.

3.2.1 Social, physical and emotional impact of Perthes' Disease

A mixed-methods study by Leo et al aimed to explore the impact that Perthes' Disease has on the social, physical and emotional wellbeing of children and their families [159]. This was published in 2020 and combined the qualitative study with a Delphi study to reach consensus regarding outcomes to be used in trials for Perthes' Disease. A survey was completed by 12 children from a UK NHS setting as part of their routine orthopaedic appointment, they completed the survey with assistance from parents. It used "emojis" through which children rated how 'happy' or 'sad' they were on a typical 'good' or 'bad' day with Perthes' Disease. The happy or sad faces related to how much they could do (happy) or if a certain aspect of Perthes' Disease stopped them doing the activity (sad). The authors reported that children identified pain as a key limiting factor to many activities including school and playing with friends, and that this was even limited on a 'good day'.

Semi-structured interviews were then carried out with 18 parents regarding the social, physical and emotional impact of Perthes' Disease. Thematic analysis demonstrated themes that impacted the child, the parents and the wider family. These included a lack of awareness of the condition and long-term outcomes from the parents, family dynamics including jealousy of siblings, and social implications such as reduced school attendance and impact on the hobbies such as sport and/or recreational activities.

Understandably there are some limitations, for example, parents assisting in the completion of the survey with younger participants could influence results. However, the study does provide some insight into the child's perspective of symptoms and their impact. Another limitation is that the participants were recruited from a single site and therefore potentially lack diversity meaning they are not representative of the patient population and could affect generalisability. A further limitation in the study is that participants were limited to mother or father as the 'family'

representative. Whilst mothers and fathers are the most common family member to bring a child to a hospital appointment, it is not the only 'type' of family member that can be impacted by Perthes' Disease. This shortcoming is a result of the recruitment strategy in which participants were recruited from their routine clinic appointment. That being said, having the viewpoint of other family members that regularly care for children with Perthes' Disease should not be overlooked. For example, a grandparent that may be responsible for children in the hours after school where activity levels are high, and activity modification may be needed most to manage pain. Notwithstanding these limitations, the study demonstrated the negative impact that Perthes' Disease has on the child and the family. It also provided a solid foundation for identifying the domains that were most important to children with Perthes' Disease and their families, which the author recognised was a vital step in the development of a core outcome set [160].

3.2.2 Understanding children and family perspectives in children's orthopaedic settings

In 2022, a qualitative study looking at the factors that influence the perspectives of children with Cerebral Palsy, their families, and the healthcare professionals caring for them [161]. The focus was regarding the children's outcomes following lower limb surgery. Similar to qualitative study, the study conducted interviews with children as well as adults. Purposive sampling was used, and the authors describe the use of a sampling matrix to ensure that the participants were as representative as possible. There were some limitations to the methodology in that the study only recruited participants from one NHS site, which poses a risk to the generalisability of the results. The authors concluded that including children and family in the planning of the management of the condition can lead to informed decision making and better outcomes. In relation to the study being reported in this chapter, it is important to recognise that the participants included those affected by the condition, as well as caregivers (both familial and clinical). Whilst the results of the study may not be generalisable, it is reasonable to suggest that the methodology is transferable.

Similar to the study outlined above, a trial comparing Surgery or Casts for Injuries of the Epicondyle in Children's Elbows (SCIENCE) had a nested qualitative study. The study highlighted the limited understanding that clinicians sometimes have regarding the experience of the patient and families [162]. Within the SCIENCE qualitative study, interviews highlighted parents' desire to understand how to do best for the child. The children interviewed within the study were able to draw on their experiences of care. The authors used this information to suggest methods of communication for surgeons when inviting participants into their study. Methodologically, this study recruited participants from an existing randomised controlled trial, which has benefits in terms of efficiency, but also has some risks associated, such as whether their experiences of care are 'typical' of patients in that service/population. The study recruited from 15 NHS sites which has the potential to increase the generalisability of the results. A significant limitation identified by the authors was the lack of fathers present in the dataset, with most of the parents in this study being mothers; the authors suggested that more research with fathers is needed. The SCIENCE study produced meaningful results relating to children's care experiences by interviewing children. This is something that has not been included in previous qualitative research in children's orthopaedics, and never with children with Perthes' Disease.

3.3 Research aims and objectives

Below are the aims and objectives of this qualitative study, which outline how the study contributes to the overall aim of this PhD programme of work.

3.3.1 Aim

a) To investigate the experiences and understanding of children/families and clinicians when considering non-surgical treatments of Perthes' Disease.
b) To explore the potential for a digital intervention to support self-management of Perthes' Disease.

3.3.2 Objectives

a) To explore how current non-surgical treatments impact on children with Perthes' Disease and their families, including how well current non-surgical treatments meet their needs.
b) To understand the information and experiences that inform clinical decision making of those who regularly manage children with Perthes' Disease and to identify the barriers and enablers to providing non-surgical care.
c) To explore the perceptions of children, their families, and clinicians when considering the development of a digital intervention to support best non-surgical treatment of Perthes' Disease, including barriers and enablers to implementation of the intervention.

3.4 Study design

This qualitative study is underpinned by a person-based approach and is intended to inform subsequent intervention development [163-165]. The person-based approach begins by establishing the views, experiences and needs of an intervention user. In this instance, the intervention 'users' are children with Perthes' Disease, their families, and the clinicians responsible for their care.

The methods used within this study are consistent with the epistemological stance taken within the PhD project, which is described in more detail in Chapter one. To summarise, the epistemological stance in this study is pragmatism [41, 42]. In the context of health-research, pragmatism supports the use of the most appropriate methods needed to address the aims and objectives identified [38]. An interview study with key stakeholders, analysed using the Framework Method enabled a synthesis of the results that informed the next stage of intervention development.

3.4.1 Theoretical underpinning

The theories that underpin this doctoral project, discussed in Chapter two, are integrated to the approach taken for this qualitative study. SDT focuses on three key elements that influence motivation: autonomy, competence and relatedness [78]. In order to understand important elements of care for children with Perthes' Disease, key stakeholders were interviewed. The study was designed to collect data that related to the autonomy of the children with Perthes' Disease and their families, by asking questions about their experiences of care including self-management approaches. There were also questions for children, families and clinicians about what future care should involve in order to satisfy these elements in an attempt to maximise motivation, and lead to a sustained change in behaviour, i.e., engagement with self-management.

In this study, as in the project as a whole, SEM supports SDT. SEM seeks to explain changes in behaviour at the level of the individuals [109]. In context to this qualitative study, it was possible to include topics for discussion related to when/where elements of care had taken place. For example, how involved family members, or external sources such as school-support staff may have influenced care for children with Perthes' Disease. The importance of this moving forward was to consider these parties in the intervention development process.

3.4.2 Methodological approach

The methods were selected to best target the study aims and objectives, and the overarching project. Qualitative interviews with children with Perthes' Disease, their families and clinicians treating the condition were conducted and then analysed using the Framework Method [119]. Qualitative interviews with participants have been effective in describing the experience of key stakeholders in similar populations, as described in section 3.2.1. The semi-structured interview methods used in this study are similar to those previously used in studies including interviews with children/parent (family) pairs [166, 167]. Interviews provide an environment in which the researcher can build rapport with the participant [153]. This can lead to the

participant feeling comfortable enough to share experiences with the interviewer that they may not have done if alternative research methods were used. This is particularly pertinent to this study, in which experiences of care are discussed, and as the patient and public involvement (PPI) activity has demonstrated, there are elements of care that are sub-optimal.

There are other methods of analysis that could have been used instead of using the Framework Method. It would be unrealistic to consider all, but two commonly used that were not chosen here, grounded theory and ethnography [168]. Grounded theory aims to develop concepts or theories based on what exists within the study data [169]. This was not the aim of this study, and therefore the method was not optimal. Similarly, ethnography relies on immersion in the setting of the population that are being studied [170]. The actions of participants in their setting were not necessary in this study, and those results would not have addressed the aims and objectives of the study.

3.4.2.1 Data collection method

Interviews were selected for this study as the data collection method, and took place with child/family pairs, hereby referred to as dyads, and clinician participants in a one to one format. Interview topic guides were designed prior to the start of the study based on PPI activities. They were designed to ensure that the aims and objectives of the study were met, but also so that participants were asked questions that allowed them to expand and share experiences that were not considered by the interviewer. A widely recognised strength of this data collection method is that it allows the researcher to ask questions on their chosen topics, gathering information in a somewhat deductive manner [171]. It also makes allowances for new themes and topics to be discussed as participants become comfortable and share elements of their lived-experience that cannot be pre-empted by a researcher.

3.4.2.2 Data analysis method

The Framework Method was used to analyse the qualitative data [111]. This method has been discussed in more detail in Chapter two. Using this method, interview transcripts were coded iteratively, with preliminary codes revised in light of coding of subsequent transcripts and applied to all interviews. Transcripts were coded, with the first five child/family and five clinician interviews coded independently by a second member of the research team (SP), who was an experienced child-health qualitative researcher. They were also checked for agreement on emerging codes. Inconsistencies in coding were discussed, and agreement reached on the subsequent coding which were reapplied to earlier transcripts. Consistent with a framework approach, some coding was deductive. Deductive analysis allows the researcher to apply their analysis based on pre-existing theory [172].

In the context of this qualitative study, the researcher followed the structure and questions included on the topic guide. These were informed by the theoretical underpinnings described earlier. An inductive approach was used, albeit less frequently, to identify concepts emerging directly from the data. Using an inductive analysis method, the researcher moved from data to theory as opposed to applying analysis to the pre-existing theory [122]. To give context to this study, the researcher used inductive analysis for new themes/topics that came about organically in the data collection. Salient themes and concepts were identified through thematic coding [125].

3.5 Sample and setting

Participants were children with Perthes' Disease, and their family, receiving treatment in one of three NHS hospitals (Leeds Children's Hospital, Alder Hey Children's Hospital and Hull University Teaching Hospitals). Recruitment from different centres was important to ensure the sample is representative of the wider UK patient population. Participants were recruited from their usual orthopaedic appointments in which they are regularly assessed by an orthopaedic consultant. Patients were sampled purposively to maximise heterogeneity e.g. differing sex, age,

treatment type (surgical/non-surgical), duration of living with condition and disease severity [173]. The children/family recruited were initially identified by a lead-clinician within each centre who was familiar with the study eligibility criteria.

Clinician participants were those managing Perthes' Disease regularly and consisted of children's orthopaedic specialists including surgeons, physiotherapists and clinical nurse specialists (CNS). Participants were sampled from a range of NHS centres within the UK (i.e. not just the three hospitals recruiting children to the study). This was done to ensure the results were generalisable and broadly reflecting practice within the NHS. There were often only a small number of clinicians specialising in Perthes' Disease found in each NHS centre.

3.5.1 Eligibility criteria

Families were eligible for inclusion if:

1) The child had been diagnosed with Perthes' Disease between one and five years ago (this is to ensure that the experience of the dyad is adequate in terms of exposure to usual NHS care).
2) Child with Perthes' Disease between 5 and 16 years old.

Families were excluded if:

1) The parent and child were unable to communicate verbally in English or engage in the interview. If the child could not communicate, but the parent wished to take part, the family remained eligible for inclusion.

Clinicians were included if:

1) They currently managed children with Perthes' Disease in their routine clinical care.
2) They had at least two years' experience of treating children with Perthes' Disease to ensure they have an understanding of usual care for this patient group.

3.6 Recruitment

Different methods of recruitment were used for child/family dyads and clinical participants.

3.6.1 Child/family identification

Recruitment of child/family dyads took place in the clinical setting, with clinicians in each centre trained regarding the study eligibility criteria and recruitment processes. On identification of a potential participant, the treating clinician (site PI) briefly outlined the purpose of the study. Any child/family dyad interested in hearing more about the study was asked for permission to share their name, email address and phone number with the interviewer. These details were shared using secure email (NHS.net) and once recruited, emails were securely stored in password protected files on University of Leeds servers and deleted from the NHS.net email inbox.

Provision of contact details did not constitute the child/family dyad agreeing to be included in the study. This was strictly used for contact by the research team. The permission to contact the child/family dyad was recorded by the PI in the patient's medical notes.

Following identification of an eligible family by the PI at a site, an email was sent from the interviewer to potential participants with the participant information pack that included an information sheet for the parent and a separate age-specific information sheet for the child (Appendix B – Child PIS and Appendix C – Family PIS). In line with current Health Research Authority (HRA) guidance, families had at least 24 hours to consider the information before the interviewer contacted them to discuss study participation. However, if a family responded sooner than this, then this was accommodated [174]. If the family had not responded within one week, a second email was sent; there was no further attempt to contact after this.

In order to provide a thorough description of the recruitment process, anonymised screening data on number of child/family dyads approached/agreed was collected by the PI at each site. Site PIs collected this as well as age (year), sex and time since

diagnosis (months). Recruitment was monitored based on the characteristics outlined above to ensure the sample was broadly representative of the typical new patient population according to both literature and the research team's clinical experience.

3.6.2 Clinician participants

For recruitment of clinicians, participation was advertised on the social media pages of orthopaedic/musculoskeletal societies or opinion leaders and further invites were sent to orthopaedic centres in the UK. The email invited clinicians that met the eligibility criteria above to take part in the study. The email contained a participant information sheet (Appendix D), outlining the aim of the study and their involvement. Responses were collected and clinicians who opted in were contacted to begin the consent process. Recruitment data, such as professional background and geographical location, was collected to ensure representation from the relevant disciplines was broad.

3.7 Consent

Informed consent was gained for all participants. Given the involvement of children under 16 in the study, a parent/legal guardian was required to provide consent for their child. This was combined with gaining verbal assent (where appropriate) from the child in the interview to demonstrate their agreement to take part in the study. The details for participant groups are detailed below and flow charts outlining the recruitment process for parents and children and clinicians are summarised in Figures 3 and 4 respectively.

3.7.1 Children/family dyads

A proportionate consent process was adopted, based on NHS Health Research Authority guidance and given the low risk nature of the data collected in this study

[174]. On contacting the family participant, the parent/legal guardian was asked to provide informed consent electronically via email. The family participant was asked to reply to the email and include the 'statement of agreement' that they were instructed to copy from the original email and read as follows:

> *'I have studied the information provided in the participant information pack and understand what will be required of 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 participation is voluntary, and I am free to withdraw from this study, unchallenged, at any time.'*

To confirm their agreement to take part, and to gain verbal assent from the child where possible, each participant (child and family member) was asked at the start of their interview if they were happy to continue and reminded that at any point they could withdraw. In all instances, receipt of consent was emailed to the participants. There was a plan for written consent in cases where potential participants did not have access to e-mail, however this did not occur. If this arose, a copy of the statement of agreement would be printed and sent by post to the participant.

Once the family member emailed providing consent for both themselves and the child, the interview was arranged. At the start of the interview, the child and family were reminded that the interview process can be stopped at any time. If the child were to have verbally refused to assent to the study the parent would still be able to participate, though this did not occur. Any child declining to take part in the interview would not have been asked for further information. It was made clear to the participants that their involvement in the study will remain confidential from their treating clinical team and that study participation would not affect their clinical care at the time or in the future.

3.7.2 Clinician participants

Clinicians that responded to the invitation had at least 24-hours to study the participant information pack and consider their involvement. To demonstrate

informed consent, the clinician participants were asked to reply to the initial email providing information outlining their agreement to involvement in this study. The email included the same 'statement of agreement' outlined for parent/child participants and read as follows:

> *'I have studied the information provided in the participant information pack and understand what will be required of 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 participation is voluntary, and I am free to withdraw from this study, unchallenged, at any time.'*

The email served as written consent for the clinician participants, outlining explicitly that the participant agreed to their inclusion in the study, including permission to use data collected in the way outlined in the participant information sheet. For example to reproduce anonymised quotes obtained in interviews as part of the dissemination. On return of this email, an interview date/time was given and completed as below.

It was made clear to the participants that their involvement in this study would remain confidential and any findings shared would be anonymised. Their clinical service was not informed of their involvement in the study.

3.8 Data collection

Following identification of appropriate participants and adequate consent processes, semi-structured interviews were carried out with children with Perthes' Disease and their families, and clinicians. The interviews followed an interview topic guide (Appendix E) and took place via video call. All interviews were audio-recorded and saved in password-protected files on University of Leeds servers. Topic guides were developed using a combination of sources. The theoretical approach (Self Determination Theory informed by the Socio-Ecological Model) informed the questions included in topic guides by considering both the intrinsic and extrinsic motivators when considering levels of physical activity. The guides were informed by

input from the PPI group, outlined previously, as well as the experience of the research team. Clinician participant topic guides included management of the condition, with consideration in terms of discipline-specific (medical, nursing and physiotherapeutic) differences. There was consideration to the needs of the child and delivery of care as well as discussion of the proposed digital intervention.

Figure 3.1 – Consent process for child/family dyads

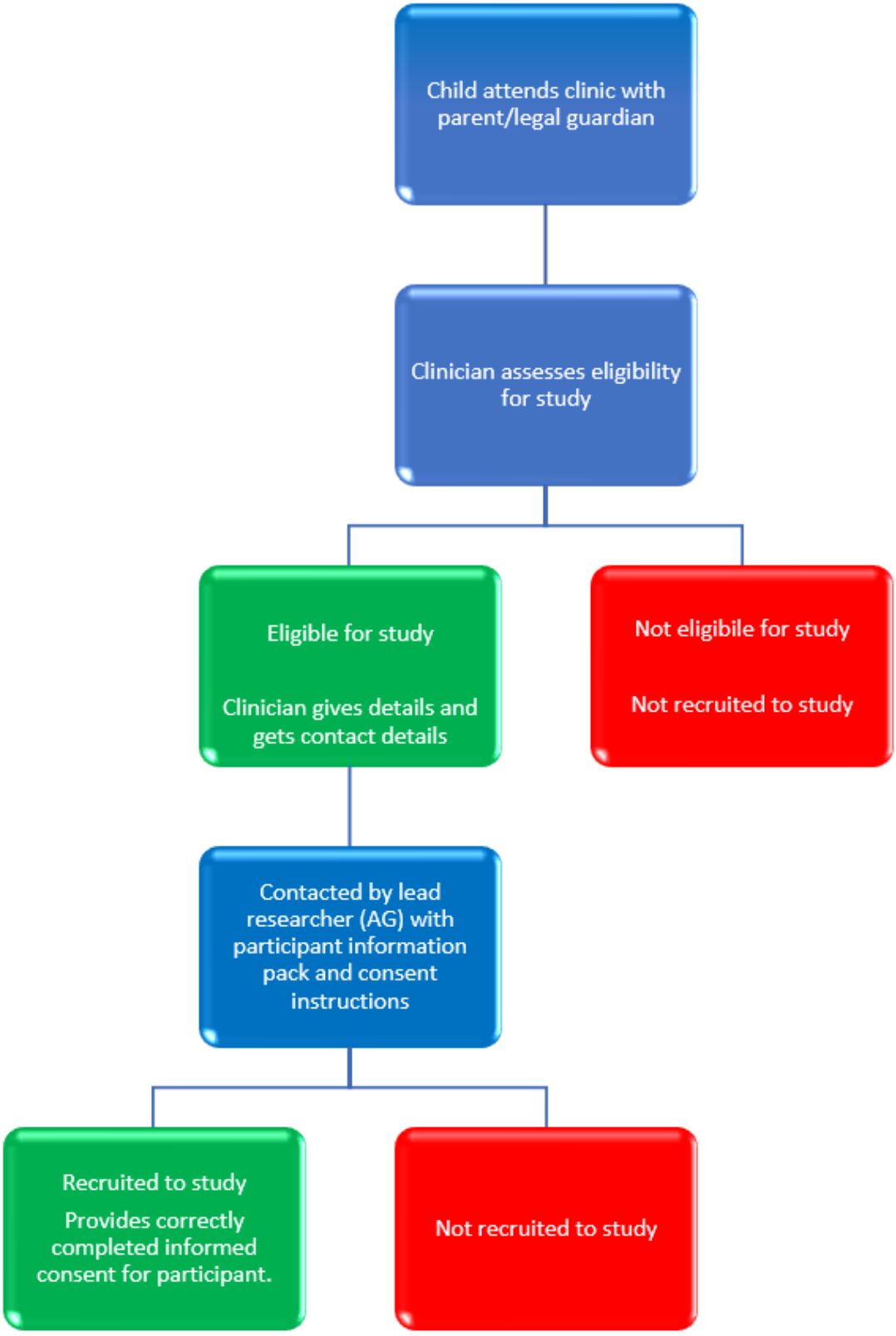
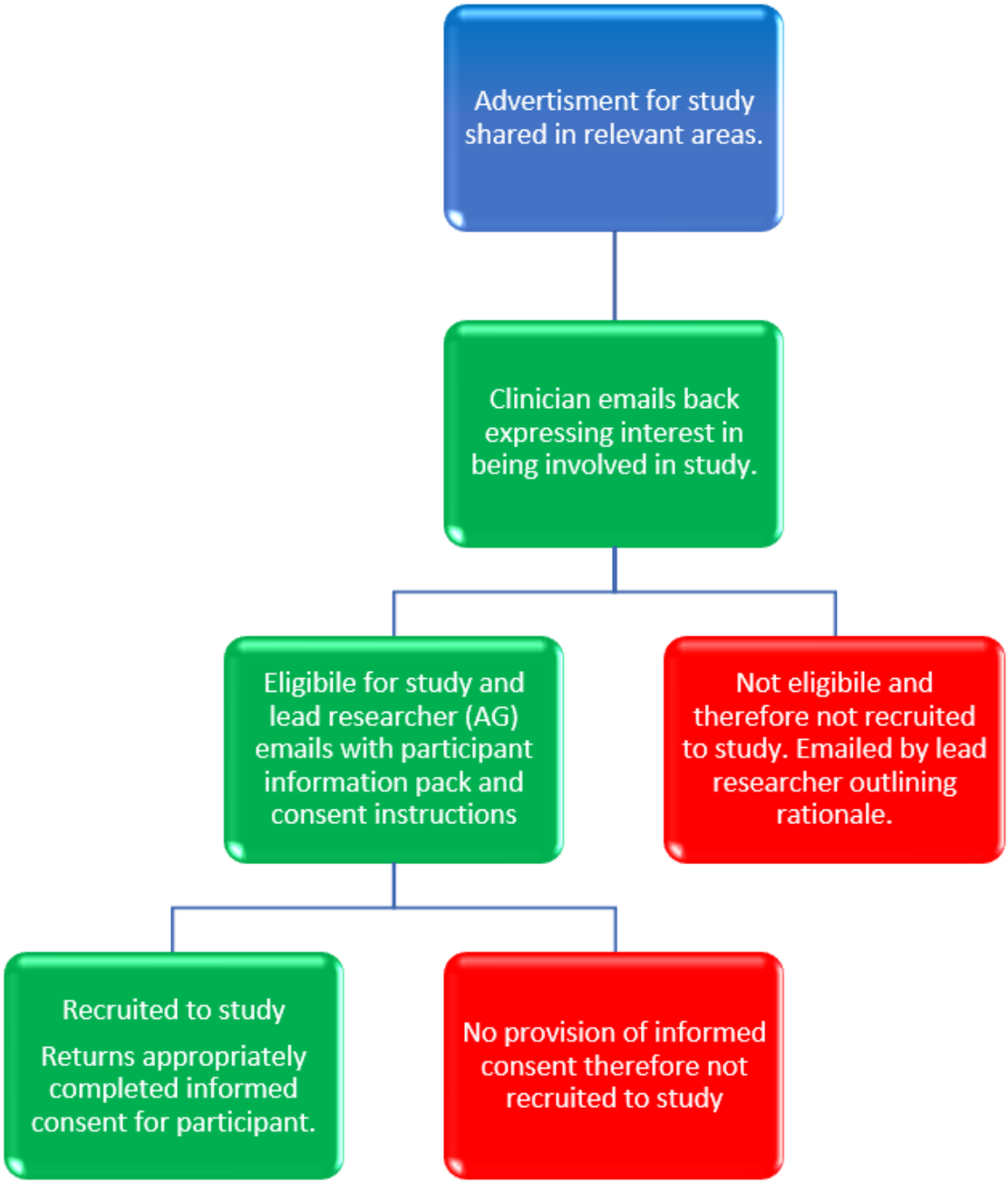


Figure 3.2 – Consent process for clinician participants



3.8.1 Interview process

After agreeing an appropriate date/time for the interview with the participant, the interview took place via video-call. Ninety-minutes was allocated for each interview. There was flexibility in this time limit to allow participants to discuss their experiences and thoughts openly in the interview. Interviews followed the relevant topic guides for the participant.

3.8.1.1 Children/family dyad interviews

When conducting an interview with a child, there was always one parent/legal guardian present. Questions to the parent/family member took place in the same interview as the child. The questions pertinent to the child were asked first to maintain as much rapport/connection with the child as possible. The questions were based on the topic guide, with the focus of this section to reflect on the experiences of clinical care offered and the degree to which it met their needs. The components of a new digital intervention were also explored and discussed here with consideration of its content and mode of delivery.

Prior to the interview starting, approximately 10-15 minutes were spent building rapport with the child. This was done by having discussions about things that interested them and in situations where needed, this included asking the child to show them something they like to do. An example of this was one child that was keen to demonstrate some moves learned in a recent martial arts session. Building this into the interview time was important to make the child feel comfortable and engaged. This time was in addition to the allotted time for the interview with the child and family member (30-90 mins). The allotted time for these interviews allowed for participants that had more or less to say. For example, if after 60 minutes a parent felt they had more to say, it would have been unreasonable to stop the interview. In the same way it would have been unrealistic to continue after 30 mins if all questions had been answered and the participants were happy that they had said all they wished to say. This was in line with previously successful methods when interviewing

children [166, 167] and aimed to make the child feel more comfortable in the interview.

In order to give the child the opportunity to openly discuss their experiences, it was important to consider that children occasionally need prompts or 'translations' from adults. This was allowed, and clearly outlined prior to the start of the interview. It was explained to family members that they were allowed to prompt and explain questions, but not to answer questions instead of the child. This was well received, and there were no observed instances of children not being able to share their thoughts. In an attempt to give the family member the same opportunity to share honest experiences, they were asked whether they would like the child to remain present during the interview. The option of having the child leave, where possible, was to maximise engagement and was taken up by some family members.

The child/family dyads were regularly asked if they were happy with the progress of the interview. It was made clear that if at any point during the interview the child became upset/does not want to take part further, the interview would be stopped, and appropriate support given. This was not the case in the majority of the interviews, however in one interview there was a particularly non-verbal child, in which attempts were made to continue discussion. This was not possible, and the interview was stopped for that particular child and the family member was asked their questions. Whilst the child did not answer all questions, they still provided answers to the early interview questions and the family member provided answers to all questions. The data, therefore, was included in the analysis.

3.8.1.2 Clinicians

Following agreement to be involved in the study, interviews with clinicians took place via video call. The interviews were based on the topic guide (Appendix E) which included a focus on the clinical decision-making process when managing children with Perthes' Disease. These topic guides were designed based on areas that are of interest but also had input from a project advisory group including children with Perthes' Disease and their family, as well as clinicians treating Perthes' Disease. This

was to ensure that the scope of the interview questions was sufficient to address the study aims and objectives, including a particular focus on barriers/enablers to providing non-surgical treatment of Perthes' Disease, as well as the content/delivery mode of a potential novel digital intervention.

Interviews considered things such as which as aspects of non-surgical care have been useful/successful in the past and if so/not, why this has been the case. Including physiotherapy, activity modification and education. The discussion around the digital intervention aimed to inform its future development. Through this, clinicians with expertise in this patient population, provided vital input on the content and delivery of a new potential intervention. All staff were asked about topics such as acceptance and usage of a proposed new digital intervention within the patient population. In addition to this, staff from different clinical backgrounds were asked their thoughts on discipline-specific clinical information/guidance. This included domains such as education for children/families on the condition and medical guidance from all staff, or for example, physiotherapy guidance from relevant staff. There were questions around how the digital intervention could be introduced into clinical practice, and utilised as a supplement to existing care to promote and empower self-management.

3.9 Data analysis

Interviews were audio-recorded, and were transcribed and anonymised, with the transcript data stored using NVivo software (Version 12, March 2020) [175] where the coding framework was applied.

Using the stages of Framework Analysis method, a provisional analytic framework was prepared prior to the commencement of interviews to support deductive coding when analysing the interviews [111]. After conducting three interviews with clinician participants, reviewing the transcripts and reading multiple times to familiarise oneself with the data, a discussion within the research team was conducted. The team included a specialist in qualitative methods (SR) who then went on to apply the initial analytic framework to the first three clinician interviews by independently

coding the data. Indexing included changes to the main themes and sub-themes within the framework matrix. These were made following discussions in the research team. For example, when charting the date, the main theme (code 5) entitled “who/when/where” was initially called “place and space” in preliminary coding, but it was agreed that “who/when/where” better captured the users of the proposed digital intervention. The final iteration of the analytic framework was then re-applied iteratively to the first three interviews, and applied to the remaining interviews within the dataset. Iterative coding was employed for the remaining interviews. Whenever minor adjustments to sub-themes were identified in later interviews, the coding was changed accordingly.

The initial framework developed for the clinician participants was found to be broadly applicable to the child/family participants. To finalise the coding for this sub-group, descriptions for child/family participants were written, applied to the first three interviews and then reviewed with the supervisory team to assess suitability. For this subsection of participants, the expertise of a different coder was utilised. The additional coder (SP) is a specialist in qualitative research in children and the discussion at this framework phase highlighted two additional codes. The codes included points raised about the ability for children and families to access any digital intervention at home, taking into consideration access to smart devices and the internet. The second code discussed clinical uncertainty for this participant type. Mostly this was in relation to the lack of strong evidence for family members on how best to manage children with Perthes’ Disease. This step within framework analysis is imperative in the process of analysis, and leads to the development of a working analytic framework that can be seen in Table 3.1 below.

A sixth code was identified over the course of the data collection, and was termed “any other business” in the first instance. This included data that did not naturally sit within the deductive coding framework. The code gathered enough data for inclusion and reflected one topic/theme – COVID 19. This code was developed using inductive coding given its prevalence and relevance within the data.

Table 3.1 – Analytic framework developed to support coding

Main theme	Notes/ideas	Clinician description (sub-themes)	Child description (sub-themes)	Family description (sub-themes)
1. Variation of care	Different approaches in different settings/trusts i.e., clinical uncertainty Needing/ wanting guidance regarding decision making	- Usual care currently at their clinical workplace - Clinicians report differences in approaches where they have worked or even changes within their own practice. - Variation of what defines 'conservative' or 'non-surgical' - Variation within 'surgical/surgeon' management. - Similar to variation, but more focussed on lack of evidence to support the clinical decision making and possibly leading to uncertainty of impact. Also need for consensus.	- If anything comes up about different experiences with different doctors or speaking to people with Perthes' that have had different care - Similar to above, not likely to come up with children but could be around the lack of knowledge.	- Comments around management of other children that met with Perthes' - Different approaches that have been used by different specialists previously seen. - Family wanting guidance and some reliable info for decisions on treatment

2. Assessing patient outcomes	Patient reported and clinical	- Debate about what a good outcome is – short/long term outcomes. - Points at which outcomes of patients are discussed, either previous patients treated with/without surgery, or prospective patients (when considering important treatment aims).	- Child demonstrating an understanding of what their outcomes are/might be. Can be long term (good hip shape) or short term (reduced pain/no surgery).	- Mentions of short (pain, function) or long term (surgery as adult, hip condition by end of disease process) outcomes for patient
3. Reason for app use	Any needs met to improve app use	- Points raised about the approach to increasing app use such as motivation (levels/rewards), exercise-related satisfaction (SDT), empowerment of patients and families. - Need to do something rather than nothing.	- Any mention of app content/previously successful approaches with apps like levels/rewards or positive experiences.	- Mentions of being told to "watch and wait" that fit with doing something rather than nothing. - SDT; motivation, family demonstrating understanding of the need for motivation in an app
4. Core features of an app	In terms of app content	- Comments about the need for consistent educational resources for patients/families outside of consultation from clinicians as well as patient-	- More awareness around what the cause of the disease is. - SDT; competency, have used apps in the past that	- Comments around variation of information available in public forums, etc. and the concern/worry that comes with this.

		sharing experience/self-education. - SDT; competency – establishment of intervention and maintenance of the behaviour. - SDT; Autonomy – ability of children to self-manage their condition	have had levels and seen improvements with this. - SDT; relatedness, to be able to discuss with other boys/girls with Perthes' Disease.	- SDT; relatedness can be where families want to be able to talk to others like themselves (mothers/fathers of Perthes' patients for example).
5. "Who/when/ where"	For app delivery	- Who the app would be used by (parents, younger or older children). - Any discussion about where, how and by whom the app would be delivered (school or parents) - Where in the care pathway the app would be introduced i.e., by a physio after diagnosis or in clinic and whether face to face, etc. - Issues around access to IT (known social deprivation issue with Perthes')	- Any discussions around use of apps at home and if not at home, then where?	- Who the app would be used by (parents, younger or older children). - Talk about using app when just diagnosed or after becoming 'experienced' patients. - Any mentions of lack of access to apps at home. - Mention of using apps/digital technologies at school - App experience of child?

6. COVID-19*	A new theme/topic that emerged from the dataset	- Any mention of COVID-19 relating to clinical services during the pandemic - Discussion around the impact of COVID-19 on patients/families with regards to their status (activity/pain levels)	- Impact of COVID-19 on child's ability to take part in activities or hobbies. - Change in Perthes' condition during pandemic (pain, stiffness, etc.)	- Impact that COVID-19 had on care provision for child/family (quality and quantity) - Experiences during pandemic which are different to experiences pre-pandemic.

*note codes 1-5 followed the deductive theoretical framework for this study. Code 6 (COVID-19) was inductive and emerged from the dataset

3.10 Ethical considerations

NHS ethics and HRA approval was obtained as the research involved NHS patients as well as family members and staff. The process outlined in the methods section above, was submitted to the NHS Research Ethics Committee (REC) and HRA and was deemed appropriate. It was awarded a favourable opinion by NHS West of Scotland REC 1 01/12/2021. REC reference 21/WS/0138, letter attached (Appendix F). This approval is in line with the most recent regulatory changes to doctoral student research [176]. Sponsorship was provided by University of Leeds. Approval from additional NHS sites research and development departments was also obtained.

Conducting research that involves children requires additional ethical consideration given that the child cannot, legally, consent to take part, but will experience the inherent burden of the research study. The HRA clearly states that whilst there is no law regarding consent of children taking part in research, that the same rules as for medical treatment are followed i.e., the child is involved in the decision making but legally, the responsibility is that of the parent/legal guardian [177].

Assent is somewhat debated in the literature, and there is a lack of consensus regarding the age at which a child can give consent or assent, and/or whether the child can provide assent at all. This is discussed in more detail below. One available definition of assent in child research is “a term to describe the child’s willingness to take part in research” [178]. In 2011, Baines highlighted that whilst UK, European and USA guidelines all outline the need for assent from children, the guidelines do not clearly state what assent includes and therefore what it means [179]. Baines concludes by suggesting that the concept of assent is sub-optimal and that competent children should be permitted to give consent, and the parents/legal guardians of children lacking the ability to consent, should discuss participation in research with those children and provide consent if deemed appropriate. In 2014, Waligora et al suggested that an age threshold for the assent process would benefit researchers, guardians and children [180]. The rationale around this was supported by literature that outlined an increase in child capacity as they grow older, however there are still issues around whether this translates to a more generalised

competence to take part in research. The suggestion of a school-age threshold for needing assent rather than consent from legal guardians seems to have been proposed in 2014 but not taken on by national regulatory bodies, such as HRA.

The email consent process used in this study is not the typical method for gaining consent, which is commonly written. However, the interviews took place remotely via video, so no face-to-face meetings took place for written consent to have been used. Sending forms out to sign also would have been less efficient in terms of time and resources. There are also practical benefits, for example, no risk of written consent forms not being returned in a timely manner which can affect data collection. With regards to assent, this was completed at the beginning of the interview with the child verbally, to ensure that they were happy to take part in the interview, a similar process also took place with the parent/legal guardian. They were made aware that they can stop the interview, unchallenged, at any point. Clinician participants provided consent using the same, proportionate process, using a statement of agreement via email prior to the arrangement of the interview.

There is a certain degree of inconvenience to consider with regards to participant time. This was considered in the methods and minimised where possible, for example, child/family dyads were recruited from pre-existing orthopaedic appointments as per their usual care. There was provision for childcare cover should any participants express their inability to take part due to lack of childcare, though this situation did not arise. Interviews were offered via video at the convenience of participants, to minimise burden on personal commitments or clinical schedules.

The research team did not identify any potential for serious adverse events (SAE) arising from participation in a one-off interview study. However, in the interest of maintaining the duty of care to the children and their families, a plan was included in the protocol and discussed with the Principal Investigators (PIs) of the sites included. In the event of any safeguarding issues coming to light when interviewing child/family dyads, the interviewer would escalate to the PI at the local site who would then follow their local policy. This process was explained to the PIs in the study set up and any PIs were given the option to discuss any issues/queries. However, due to the close

working of each of the PIs there were no anticipated issues regarding communicating any escalation of safeguarding concerns and thankfully, none arose.

3.11 Results

An overview of each of the six main themes is provided, with further detail given for the sub-themes that emerged within each main theme (Table 3.1). The first five follow the analytic framework outlined in previous sections. A sixth main theme included in this section, refers to a recurrent theme identified in the analysis process, which was COVID-19. Illustrative participant quotes are used to give context and support the main themes and sub-themes identified within the dataset. A quote table, including the fuller dataset is provided (Appendix G). A thematic table displaying the frequency of each theme mentioned by the participants is also provided (Appendix H).

The characteristics of the sample are displayed in Table 3.2 below. In the interest of maintaining participant anonymity, in the clinician participant group, the professional role has been given but nothing else. For the child/family participant group, children aged 6-11 years old are described as “young” children and 12-16 years old as “older” children.

Table 3.2 – Characteristics of participants in the study

Characteristic	**Child**	**Family**	**Clinician**
Participants, n	12	12	12
Female, n (%)	3 (25)	11* (92)	9 (75)
Age, years, mean (range)	10.8 (6 to 16)	N/A	N/A
Previous surgery, n (%)	6 (50)	N/A	N/A
Physiotherapist, n (%)	N/A	N/A	6 (50)
Surgeon, n (%)	N/A	N/A	5 (42)
Clinical Nurse Specialist, n (%)	N/A	N/A	1 (8)

*10 mothers and 1 grandmother. Remaining family member was a father.

3.11.1 Theme one: Variation of care

Widespread variation in care of children with Perthes' Disease is well documented [28]. In an attempt to understand this better, this theme explores the experiences and perspectives of key stakeholders with regards to this variation. The sub-themes within this are 'current usual care'; 'different approaches'; 'evidence to support decision making' and 'agreement amongst clinicians.'

Current usual care

The first sub-theme focuses on the care that currently exists in their Perthes' Disease treatment. Amongst clinicians, this focussed on their clinical practice, and for children and families with Perthes' Disease, this focussed on current treatment. In this sub-theme, participants described variation in terms of current management approaches for Perthes' Disease and elements of clinical treatment. There were, however, terms that were used regularly in the interviews, particularly within the clinician participants. The term "normal life" was used multiple times in the individual interviews, it was used as a reason for the treatment approaches clinicians used including things like physiotherapy for range of motion exercises, pain control with/without analgesia and activity modification. One surgeon explained their treatment approach:

> *"my mainstay of treatment is to maintain their range of motion, make sure that their pain is controlled, and to let them have as normal life as possible"*
>
> *Clinician 1, Surgeon*

Activity modification was something that was mentioned in many interviews, and when discussing the current care children with Perthes' Disease receive, many participants talked about limiting children from high-impact activities. This is thought to be fairly common practice; however, it was generally reported that activity modification required the understanding and involvement of the family. In an attempt to preserve a "normal life", it was often the most extreme forms of 'high-impact' activities that were advised against:

"our approach is to avoid bouncy castles and trampolines, but otherwise let them have a normal, a normal life"

Clinician 8, Surgeon

Physiotherapy including hydrotherapy was regularly discussed, often as an alternative to the high-impact activities outlined above. A clinician explained:

"If they've got a decreased range of abduction then I refer them to physio and ask the physio to assess them for hydro if they have that available to them".

Clinician 7, Surgeon

Child/family and clinician participants discussed the benefits of water-based exercise; whether in formal physiotherapy sessions or extra-curricular. When asked what they like about their physiotherapy treatment for Perthes' Disease, child 1, 9 year-old female reported "I get to swim more".

Access was mentioned numerous times, as seen in the discussion with the clinician participant above. Clinician and family participants stated how effective water-based therapy can be, but many participants highlighted that their hydrotherapy pool had closed, or access had been removed. Some clinician participants highlighted the lack of physiotherapy access in their clinical service and the potential service-related risks that are associated with that. Whilst this was only present in a minority of interviews, this is important to consider in the preparation of a self-management intervention.

<u>Different approaches</u>

The second sub-theme builds on the first by exploring the different approaches and treatments that participants have experienced for Perthes' Disease. For clinicians this involved a range of different working experiences, often from different geographical areas they had worked, and for children and families, explanations of previous care within their "Perthes' journey" were explored.

Participants of all kinds drew on their experience of different approaches used for Perthes' Disease in this second sub-theme. Many clinician participants explained

variation was because of the lack of evidence to support a specific treatment approach. Often care was subjective and based on the experience of the clinician during training, commonly in different geographical locations. Children and families recounted having different opinions from clinical teams in the NHS services, compared to private practice, where they were told to be "100% non-weightbearing". Others talked about different treatments they've tried in the past including non-NHS, self-directed approaches such as reflexology and massage.

Particularly prevalent within this sub-theme was the inconsistency in terms of sometimes opting for operative care, and in other instances opting for non-operative care. In particular, one surgeon expressed the uncertainty around the approaches used:

> *"In the early years in my practice, when I was quite keen, I probably operated on more than I would now. And I have a suspicion that they're the ones that had the good outcomes, so they're probably the ones that if I left alone would probably have done quite well as well."*
>
> *Clinician 2, Surgeon*

Surgical interventions such as total hip replacement (THR), that had previously been seen as 'last-resort' were also discussed. With a number of clinicians explaining that this non-typical approach had been used in severe cases and led to good outcomes. With one surgeon reporting:

> *"as orthopaedic surgeons our whole career we've been told, no, no, no, you can't do an early hip replacement, you have to wait until they can't walk anymore and in terrible pain. But actually younger people do very well, and it gives them a new lease of life, so it's not the worst outcome in the world if that's what they end up having"*
>
> *Clinician 1, Surgeon*

Clinicians explained that over time, non-surgical treatment has become more prevalent, but previously there were things such as bracing, casting and "slings and springs" used for children with Perthes' Disease. The amount of multi-disciplinary

team (MDT) support also seemed to affect decision making previously, with some clinicians explaining that choosing a conservative approach was easier when regular physiotherapy was available.

Similarly, some clinicians explained:

> *"when I first started work, all the information was 'no impact' (referring to impact on the hip)"*
>
> *Clinician 9, Physiotherapist*

However, many explained that with more regular monitoring of things like range of movement and activity levels, this approach is not as common.

Evidence to support decision making

The third sub-theme explored participants' understanding of the available evidence to support the decision-making process for the approaches used previously or currently. Given the nature of Perthes' Disease and the lack of robust evidence in the area, this often-involved clinicians referring to a lack of evidence to support treatment approaches. Clinicians particularly outlined treatment approaches that they had used, even uncommon treatments, such as steroid injection and early total hip replacement. Whilst clinicians identified that these were effective at the time, they were unsure of the long-term effects. This uncertainty was evident across participants, one participant summed up the desire for more guidance clearly:

> *"you see a child with Perthes', and you genuinely don't really know in your heart of hearts the best treatment algorithm for them"*
>
> *Clinician 7, Surgeon*

In line with this, participants discussed the need for clinical consensus to guide treatment, and provide the evidence base for treatment. A handful of participants referred to one or two studies that have provided recommendations for surgical intervention. This was, however, often followed up with a concern raised about the outcomes used in the studies. Clinician participants explained that outcomes in

studies were often radiological and didn't capture "functional outcomes" for patients. When discussing surgical intervention impacting children with Perthes' Disease, a participant reported:

> *"even if it didn't make a difference to the hip, so, to the shape of the hip which is what surgeons care about, I'd be interested to see whether surgery made the difference to patient outcomes. To, kind of, pain and stuff. Because I don't know that"*
>
> *Clinician 8, Surgeon*

The uncertainty was also present in child and family interviews. Children expressed their desire to know more about the condition in the long-term, and described the potential benefit of knowing more:

> *"if you know how long you're going to have it for you will be like me, I'm just…I don't know, I'm just waiting for it to go, if you know when it's going to go probably, you're waiting for your best day of your life basically"*
>
> *Child 7, young male*

<u>Agreement amongst clinicians</u>

In the final sub-theme, participants discussed their experiences and exposure to clinicians and the common trend of disagreement between them with regards to treatment approaches. As outlined in previous sub-themes, variation exists, and participants discussed the range of treatment options leading to a distinct lack of agreement between clinicians. One participant expressed their disagreement with a previously experienced approach:

> *"I certainly didn't agree with the ethos of let's put them in a wheelchair for a year, because I don't think that made any difference to the outcome"*
>
> *Clinician 2, Surgeon*

The disagreement amongst clinicians was even recalled by the families of children with Perthes' Disease. One mother described a discussion with a surgeon:

“that's what (my consultant) said. He was like, I'm so sorry, if you go and see any consultant, we’ll all say something different”

Family member 7, Mother of young male

The clinician participants highlighted the need for this to change, and mentioned the need for consensus on treatment options on a number of occasions. With some participants calling for the inclusion of national bodies to be involved with attaining clinical consensus. To ensure that children with Perthes’ Disease receive the optimal care and that clinicians are “all giving the same kind of information”.

3.11.2 Theme two: Assessing patient outcomes

Understanding outcomes that are important to key stakeholders is vital to understanding the care undertaken so far. It also helps inform any future interventions that may be developed and implemented. Furthermore, agreement about key outcomes ensures that comparisons between treatment choices can be made in a formal sense. The sub-themes are ‘defining outcomes’ and ‘rationale for treatment approach’.

Defining outcomes

Clinicians and children/families alike all have desired outcomes in their mind when considering treatment approaches. Of course, the decision making behind this may differ based on many factors such as experience, knowledge or priorities, nevertheless, the decisions are based on outcomes. To explore the outcomes of importance to key stakeholders, this sub-theme demonstrates points at which participants discussed any outcomes for children with Perthes’ Disease, including clinical outcomes such as radiological outcomes or need for surgery as an adult, or more functional outcomes such as being able to take part in physical activity.

All participants (clinicians, children and family members) emphasised the importance of maintaining levels of function and minimising pain when considering treatment

approaches. Although clinical outcomes (e.g. radiological findings) were reported, the social interaction of the child was highlighted. Some participants also discussed school attendance and subsequent mental health issues.

> *"I'm thinking about the child, I don't want them to be in pain, I don't want them to be limping, I don't want them to be off school for six months so that they get mental health issues, which we're seeing a lot of right now"*
>
> *Clinician 3, Surgeon*

When considering the long term, participants repeatedly referred to the link between a poor outcome and the early need for total hip replacement (THR). It was interesting to hear both clinician and child/family participants report that early THR was not necessarily a bad outcome, if it resulted in significant improvements in function, pain and quality of life. One family member recalled a point at which the child claimed:

> *"if it means getting rid of the pain, I'll have the operation and have a new hip"*
>
> *Child 2, young male*

The quote above demonstrates the impact that pain has on the child's quality of life clearly. Another point of interest that was key to the interviews was the need for more research around outcomes following intervention, including research to find out what is a 'good' or 'bad' outcome for the patient. A clinician participant (Clinician 8, Surgeon) demonstrated the lack of ability to measure what is a 'good' outcome for children, "I genuinely don't know what's successful and what's not successful".

<u>Rationale for treatment approach</u>

Participants discussed the reasons for why a treatment approach was chosen. These approaches varied depending on whether it was clinician or child/family participants, but they were largely based on the child's perceived outcomes. Whether that be short-term outcomes such as pain and function, or longer-term outcomes such as the need for early total hip replacement and hip condition at skeletal maturity. Understanding these decision-making experiences and what is important to both

clinicians and children/families can be useful for things such as the development of interventions for this patient population.

Typically participants discussed ROM, pain and functional ability as key indicators for when interventions such as surgery were needed. With many reporting that those with good ROM and minimal pain doing well with just physiotherapy and those presenting with poor ROM and more pain tend to go on to have surgical intervention. This was the case for both children having minimal input and those having relatively significant input. With 'hands-off' approaches aiming to, in the words of one participant (Clinician 8, Surgeon) "let your kid be normal". In contrast, another participant explained that whilst a THR seemed extreme, it had dramatically improved their activity levels, from what was a previously low-level state:

"*these kids are in pain, it's limiting their function…they're getting pain daily, they're limping, they're having to use a stick at university, they can't participate in sports. And actually you give them a hip replacement and they're cracking on like nothing's ever wrong and they love it. I had one boy emailed me from climbing Machu Picchu in Peru for his follow-up PROMs data and he'd had it for Perthes'.*"

Clinician 3, Surgeon

Clinicians reported that the older a child was at presentation, the worse the outcome tended to be and that it often influenced their treatment decisions. Children/family participants were also aware that age influenced their treatment options. There was, on more than one occasion, discussion that treatment approaches were not based on rationale, rather based on experience with some suggesting that any type of intervention did not impact outcome. One clinician participant said,

> *"I've got to put my hands up and say I do very little with these kids now because over time you know that these kids, a lot of them will come out the other end no matter what you do with their own outcome".*

Clinician 10, Physiotherapist

A further consideration highlighted when recommending treatments was the financial ability of families to provide some therapies, such as swimming and cycling. Whilst they are commonly advised, a physiotherapist explained the potential burden,

> "*The financial ability of the parent and their time to be able to take the kid swimming, to access swimming, to access cycles – they may not be able to afford a bike.*"
>
> *Clinician 9, Physiotherapist*

3.11.3 Theme three: Reasons for app use

Understanding the motivation for any participant to use a potential intervention is vital to the design and development process. In this study, thoughts and experiences on app use were explored, both in the context of this project i.e., for a Perthes' Disease self-management digital intervention, and in similar types of apps. The sub-themes within this theme are 'previously successful or beneficial apps'; 'doing something rather than nothing' and 'rewards and levels'.

Previously successful or beneficial apps

There are no apps for children with Perthes' Disease, of any kind. Therefore, in order to design and develop one for the self-management of the condition, participants were asked about their experiences of using apps in other areas of life. This was to explore whether aspects of success could be transferred into this context to optimise user-experience.

Clinicians were able to draw on experience from using apps in other clinical areas that they work in. A clinical nurse specialist discussed the success that an app had yielded in a limb-reconstruction clinic where children were responsible for their programme of external fixation maintenance. This participant (Clinician 11, CNS) specified that the success was linked to "the kids getting involved and doing it themselves" which any potential app for Perthes' Disease would aim to do in order to increase autonomy

and independence. Similarly, a physiotherapist reported that an app used with patients with rheumatological conditions had been successful and when discussing the potential for an app for Perthes' Disease, suggested:

> *"if we could use it in a similar way to how the rheumatology self-management app works, I think that might improve compliance"*
>
> *Clinician 9, Physiotherapist*

This has particular importance when considering previously discussed themes that outline the variation of care within Perthes' Disease. The feelings that participants in this study and previous PPI work have expressed that engagement is an issue with self-management aspects such as physiotherapy.

Using apps is not novel to children; many children explained how they have used apps in other settings e.g. school homework. A common undertone within this sub-theme was that children used apps for homework that was previously quite difficult to engage with and had even resulted in an educational benefit. This was demonstrated well by an eight-year-old child with Perthes' Disease, when asked if they liked using an online maths homework:

> *"Yes, because it's fun and it's not like normal maths homework"*
>
> *Child 1, young female*

One parent of a sixteen-year-old (Family member 6, father of older child) explained that "apps are something they're more comfortable with, so I think (an app for Perthes' Disease) is a great idea".

<u>Doing something rather than nothing</u>

This sub-theme relates to regularly reported attitude regarding the treatment of Perthes' Disease from clinicians and families generated in PPI activities leading to this study. Often non-surgical treatment of Perthes' Disease involves regular observation, which may be called 'active observation' or equivalent. This sub-theme describes responses from participants that have talked about having a self-management app

for Perthes' Disease to offer 'something rather than nothing' for these children and families.

The responses within this sub-theme were from clinicians, and all described a similar concept, empowerment. All-but confirming what is thought from the available literature, a participant described how children with Perthes' Disease and their families often feel when the decision is made to proceed with non-surgical treatment:

> *"I think that sometimes you get a diagnosis of Perthes', and if your orthopaedic surgeon is going down a nonsurgical route, I think that in one way the parents and the children are pleased they don't need the surgery, but in another way then they're not having, in their mind, an active treatment"*
>
> *Clinician 7, Surgeon*

A clinician participant (Clinician 1, Surgeon) described the app as an intervention that could make users "feel empowered that they're actually doing something to manage their own treatment". Another participant hinted towards this, stating that:

> *"an app might be a useful way for parents to engage and feel that someone cares about them"*
>
> *Clinician 6, Surgeon*

<u>Rewards and levels</u>

The final sub-theme refers to the theoretical underpinning of this study. Linking rewards and levels within a potential app could result in improved adherence by increasing things like their levels of exercise-related satisfaction, autonomy and empowerment.

An app for Perthes' Disease would need to motivate users to engage. The most likely users of the app from the responses gained in this interview study were children with Perthes' Disease and their families. The SDT, which underpins this study, links motivation to increased levels of physical activity by addressing things such as exercise-based satisfaction and autonomy. Participants in this study referred to the

need for an app to include fun and innovative ways to engage children and also suggested that rewards and levels would be effective in doing so.

There were references made by all types of participants about the potential to use children's favourite characters within an app such as video game characters or TV show characters. Similarly avatars were mentioned frequently by participants, an eight-year-old participant suggested:

> *"you could get points every time and then you can use those points to make your Avatar"*
>
> *Child 3, young male*

Children particularly, explained that they would like to design their avatar so it could look like them, and some reported having done this in previous apps. This as well as utilising children's favourite characters also links well to the SDT as the theory has frequently demonstrated a link between target populations benefiting from increased levels of relatedness. Being able to see their favourite characters, or more so a representation of themselves, has the potential to significantly increase motivation and result in increased engagement with a self-management app for Perthes' Disease. Clinicians and other family members also expressed the need for incentives for children to complete things, like exercise regimes and also mentioned that role models and rewards would optimise this.

3.11.4 Theme four: Core features of an app

This theme explored participants' thoughts around what an app should consist of. The sub-themes are 'educational resources regarding condition'; 'communication' and 'self-management within an app'.

Educational resources regarding condition

Being able to provide educational resources for children with Perthes' Disease and their families is not a new problem. The participants made that abundantly clear and

had very clear direction for what a digital intervention might include in that regard. There were frequent mentions of consistent and reliable information that could be stored in an app. Many reported that existing resources and methods of information gathering, such as social media or simple search engine reviews, can be dangerous. When asked about the use of an app for educational resources about Perthes' Disease, a mother of a 9-year-old explained:

> *"I think that'd be good, yes, if there was more information in one place (an app). One that isn't just like a scary site of mums all freaking out"*
>
> *Family member 11, mother of young female*

Having a centralised place for children with Perthes' Disease and their families to refer to had many potential benefits for participants. Clinicians, children and families mentioned the benefit of having information digitally available meaning it didn't get lost. Participants also mentioned the benefit of information being available in between hospital appointments which can often be long periods of time. Educational information was also thought to be of use in other settings, such as schools, to raise awareness and educate others on the condition.

A final element of education that arose from the interview responses was the empowerment benefit for children to have explanations that are suited to their age/ability, to explain the condition. One mother highlighted that this information may have helped her child understand his condition more, and also to understand why his activity was restricted in the early stages of the disease.

> *"He can't go and join a football team, he's not allowed to go on a bouncy castle, if he could have a bit more understanding of why he can't do those things, I don't know if that would help, but that's probably the one thing that he really, really, struggles with"*
>
> *Family member 8, mother of young male*

Communication

Being able to discuss Perthes' Disease in a range of ways was something that almost every participant highlighted in their interview responses. The most common was the potential for 'forums' or 'chat rooms' for children/families to talk to other children with Perthes' Disease/families. Understandably, there are safety concerns around this, and participants highlighted this too. Nevertheless, participants highlighted the potential benefits for children with Perthes' Disease being able to discuss "how annoying is my mum telling me no all the time?" (Family member 1, mother of young female).

Communicating with medical staff between clinic visits was also discussed by many participants, both from children/families and clinicians. Similar to the potential for inter-patient communication, it is certainly something that has risks involved and was also acknowledged by participants. One surgeon (Clinician 7, Surgeon) highlighted the benefits that communication with healthcare professionals could have, but also explained that direct communication from parents and families was "just too much", and may set unrealistic expectations regarding the clinical service's ability to respond.

Clinical markers and progress also featured regularly in this theme, with participants discussing the potential for physiotherapy regime completion being trackable and perhaps even checkable by the physiotherapy team. Also, the ability to bring 'diaries' of things like physiotherapy, sleep and activity was something that many participants thought would be a positive element in an app.

Self-management within an app

As demonstrated in the attached thematic table (Appendix H), there was at least one response from each participant relating to this sub-theme. Designing a digital intervention such as an app to enable children with Perthes' Disease and their families to self-manage the condition had benefits to all involved. Clinicians talked of the benefits of introducing an app to children and their families:

> *"giving a patient some kind of intervention so that they feel empowered that they're actually doing something to manage their own treatment or their child's treatment, I think will go down really well"*
>
> *Clinician 1, Surgeon*

This was echoed by clinicians and child/family participants alike, who referred to the need for "ownership". In the interviews, children replied that they wanted to know what to do when it hurts and what they can do themselves. A clinician participant (Clinician 9, Physiotherapist) highlighted the need for child-involvement and autonomy within their own care, irrespective of the age of the child, stating that "no matter how young the kids are, they want to be involved in their own care". Similarly, a clinical nurse specialist in orthopaedics highlighted the potential benefit of an app for children that were transitioning to adult care, when discussing the use of an app:

> *"it's encouraging them to be more autonomous in their care, because that's teenage transition. We're teaching young people to take responsibility for their care"*
>
> *Clinician 11, Clinical Nurse Specialist*

Understandably there were some reservations around the potential for self-management interventions to be clearly defined. For instance, with this potential digital self-management intervention, it would not be the case that an app would replace care. Rather, it would supplement existing care and provide an intervention that would support children with Perthes' Disease and their families over the course of the disease process. One surgeon highlighted the importance of this by reporting:

> *"Do I want an app telling what I should do for my patients? Hell no".*
>
> *Clinician 3, Surgeon*

This highlights the need for any design and development to include a tailored training plan for the users, and those providing access. This will ensure that all parties involved understand the role and scope of the intervention.

3.11.5 Theme five: Who, when, where

To explore the thoughts of key stakeholders, questions were asked relating to using an app as a self-management intervention for Perthes' Disease. The sub-themes are 'different users of the app'; 'when the app gets used' and 'where the app gets used'.

Different users of the app

Understanding who key stakeholders perceive to be the main users of any potential app is important, to ensure any future planning is based on the opinion of those that will have the app presented to them. Clinicians and family members alike both recognised very frequently that children are particularly accepting of apps. This seemed to be irrespective of age, with it never being mentioned that a child may be "too young" and one participant even reporting that "most three- and four-year olds can navigate a smartphone now". Because of this, many reported that they think children using the app is a good idea for future self-management of the condition. This was supported by numerous mentions of autonomy and "ownership" on the child's behalf with regards to the condition as shown in the quote below:

> *"The other thing I love with apps is that it enables the child to take some ownership and some responsibility, and they have therefore some understanding of what they're trying to achieve. No matter how young the kids are, they want to be involved in their own care on the whole, I've found. And I think that's important to acknowledge that and to respect that and to enable that. So yeah, bloody love an app"*
>
> *Clinician 9, Physiotherapist*

There were mentions from clinicians about the potential for socioeconomic status or deprivation to impact the accessibility of a potential app. Things such as smart-device access and mobile data both arose as a potential barrier to use, however these were significantly outweighed by the number of clinicians suggesting that on the whole most families they treated, irrespective of socioeconomic status, had access to smart-devices. Interestingly this was never mentioned in the interviews with

children/families, further supporting the likelihood of access not being an issue for children with Perthes' Disease. It is worth noting that in the sections below, there are a number of references to the fact that any difficulty to accessing an app could be addressed with things such as access in school or access in hospital using devices owned by respective organisations.

In the interviews with children/family members, it became apparent that the ability for parents to access reliable information about Perthes' Disease was important. Whilst many suggested a potential app should be largely child-facing, many also explained that an interface for parents could be of significant benefit. One parent (Family member 10, mother of older female) mentioned the benefit of having a safe place to go for information "because there's a lot of scaremongering that goes on online".

<u>When the app gets used</u>

In this sub-theme, the focus was around when the app would be used, both in terms of what time of the day the app would be used e.g., before/after school and when in the 'Perthes' journey' the app might get used. The latter refers to discussions with participants about the app being used at different time-points in the disease process, for instance, initial diagnosis, to provide information on the condition or throughout the condition for self-management activities, such as physiotherapy or access to frequently asked questions.

Children/family participants discussed the use of an app almost as if it becomes part of their routine, and one mother highlighted that if it was to support things such as physiotherapy at home, it would have a benefit for the child and parent. Stating that children with their own app could complete their advised programmes because:

> *"when we were given the physio initially it was really time-consuming for me to do. So if the child had their own app to follow themselves, it just frees up parents' time".*
>
> *Family member 4, Mother of older male*

Clinicians had more input regarding when it may be most useful in terms of the stage in the disease process. An interesting insight from a Clinical Nurse Specialist highlighted the potential for autonomy in teenagers. They highlighted that as they head towards the age at which they would transition to adult services, an app for these young-adults would be useful in enabling them to self-manage more effectively.

Another clinician participant highlighted the importance of timing the activities offered within an app carefully, suggesting that, as with some physiotherapy modalities, excessive advice to exercise may exacerbate symptoms. This surgeon explained that stretching to increase range of movement may be good in the later stages of Perthes' Disease, but in the initial stages could irritate an already inflamed hip joint.

Where the app would get used

Understanding where key stakeholders perceive patients/families would use an app for self-management of Perthes' Disease is vital to its design and development. In this sub-theme, the considerations that participants had given towards the location of app use is explored.

Many participants mentioned that an app would be of benefit because of its potential use at home, meaning things like children being able to access information and advice independently at home and complete physiotherapy exercises. A CNS explained the importance this could have on not needing to come to hospital for repeat appointments if the patient was able to self-manage using the app.

School was ever-present in the discussion of where an app might be used, with clinician participants suggesting that given the access that modern schools have to technology, any issues around access to smart devices can be eliminated by children accessing the app at a school. Similarly, it was mentioned by a clinician participant that access to smart devices could be done in a hospital setting in the clinics if access was difficult. School remained common and was brought up by a child who

mentioned that an app would help her at school from things that have the potential to impact not only physical symptoms but social issues as well:

> *"the teachers wouldn't have to do one on one exercises, I can do it in PE, when the other kids are doing activities that I can't do"*
>
> *Child 11, young female*

Being able to use an app in school was mentioned in this way to demonstrate the autonomy that could develop for a child using it.

3.11.6 Theme six: COVID impact

This theme was not part of the analytic framework, at any iteration. However, the impact of COVID is in all areas of life, and the treatment of Perthes' Disease is not unscathed. Clinician and child/family participants expressed the impact that COVID-related lockdown and restrictions had on care for Perthes' Disease. Some participants explained the negative effects, such as children not being seen regularly. Previous themes have outlined the feelings of children with Perthes' Disease and their families with regard to a lack of engagement between hospital visits. This was only exacerbated during the pandemic. A mother explained:

> *"we've had COVID for two years so we, kind of, haven't been seen. We've just been shoved on a shelf"*
>
> *Family member 1, Mother of young female*

The impact was felt by clinicians too, with pressures on waiting lists meaning some surgeons not able to carry out procedures. As highlighted earlier, low-impact activities such as hydrotherapy are common in treatment for Perthes' Disease. COVID also led to logistical issues for services:

> *"We used to use hydrotherapy, but we unfortunately don't have a pool anymore, it was closed during COVID and it's not looking like it's going to open"*
>
> *Clinician 5, Physiotherapist*

Amongst the understandable negative results of the pandemic, there was one particular benefit that participants mentioned frequently. A participant explained:

> *"silver lining of COVID is everybody's become so much more au fait with technology"*
>
> *Clinician 6, Physiotherapist*

This was recognised by clinicians and child/family participants, and it is very likely that this is a contributing factor to the positive responses for a potential digital self-management intervention for children with Perthes' Disease.

3.12 Discussion

3.12.1 Main findings

This study was the first to explore the experiences of children with Perthes' Disease, their families and the clinicians who provide treatment. Participants of all kinds described a variation in the care they deliver and receive. Clinicians particularly described that there was a lack of agreement amongst clinicians with no robust evidence to support their treatment choices. They also demonstrated the desire for more information and evidence to support treatment for Perthes' Disease so that the outcomes for the children were optimised. The general feelings from the participants were that an app would be an appropriate method of aiding self-management for non-surgical treatment of Perthes' Disease.

Variation within centres treating children with Perthes' Disease in the UK has been demonstrated previously by the researcher [28] but the factors driving variation were not explored in that study. Throughout the interviews, participants told us what the variation was, e.g. some surgeons opting for a more conservative approach as they have gained more experience. This is something that the previous study was not able to capture [28]. Similarly, the lack of agreement is something that is known, and research focuses have been directed towards finding the most effective treatment [181]**.** However, this study highlighted the desire from clinicians as well as child/family

participants for better evidence to support their decision making when considering non-surgical treatments for Perthes' Disease. Digitalisation within the NHS is already taking place, and ensuring that future treatments target the needs of those using it is imperative [182]. This study demonstrated the acceptance and positive response to a digital intervention in the self-management of Perthes' Disease. Previous PPI work has also yielded a positive response to this suggestion, but this study was able to involve key stakeholders in the first stages of design. Their comments on content, and delivery, are vital to the next stage of intervention development.

With clinical uncertainty, there are inherent risks associated with making treatment decisions. This is the first study that has given clinicians the opportunity to discuss their uncertainty in a research setting. Children with Perthes' Disease do not require regular appointments with healthcare professionals and often are living with a condition that is debilitating, with little input. The opportunity to discuss their experience is something that participants responded to well. There is a potential that the recent COVID-19 pandemic played a significant part in the desire to share experience. That being said, the desire to 'do something rather than nothing' was ever present throughout all participants. This may explain the positive reaction to self-management, as well as the insights into what will positively motivate children to engage with a digital self-management intervention.

3.12.2 Strengths and limitations

Having the patients and public that represent the population give insight into interventions in the future (the use of an app) was a real strength of this study. It meant the researcher could combine their input with theoretical underpinning such as SDT and SEM to maximise the outputs in this qualitative interview study. The iterations of the coding framework and interview schedule are examples of how the PPI input informed by the theoretical underpinning affected study design. For instance, SEM was useful when considering the 'levels' that influence care i.e. the child's independence, family support and organisational factors (whether that be school, or a clinical organisation). SDT mapped to the themes of the study with clear

demonstrations of elements such as the desire for education from both child and family participants. This aligns particularly with the 'competence' element of SDT which influences motivation.

As with any study, this qualitative piece of work was not without its limitations. Children/family dyads were sampled from three NHS sites via their existing orthopaedic appointments. Collecting data from three sites only poses a risk of not representing the heterogeneity of the patient population as a whole, which might limit the transferability of study findings. To counteract this, purposive sampling was used, as well as clear discussions and instructions to PIs at each recruiting site. This was to ensure that participants were recruited to accurately represent the diversity of the patient population, but also the demographic of family members. This worked for children, with a varied cohort of sex, age and surgical/non-surgical management previously. However, family member homogeneity was more significant. Of twelve family members, there was one grandmother, one father, and the rest were the mothers of the children. This was somewhat bound by the responses and the scope of this project as part of a PhD, for instance, five fathers were contacted to take part after expressing interest, but only one responded to the invitation. There is a potential for their viewpoint to be different to that received in this study, and could be considered in future studies. A further limitation within the study was the interview topics relating to app development. Participants were informed about the PPI activity and previous work that had led to the plan for the development of an app in the PhD project. The risk associated with this is that it could influence participant's feelings and response towards the new intervention. This was considered prior to the start of the study, and felt unavoidable given the need for input as part of the intervention development process. Although it was unavoidable in this particular study, it was something that was considered in preparation for the nested qualitative study as part of the final study in Chapter six.

Understanding the factors influencing the decision-making processes amongst clinicians treating children with Perthes' Disease has not been investigated before. Being able to provide insight of a sample from the breadth of the UK, from regional children's orthopaedic centres to smaller centres that do not treat many children with

the condition, is a strength of this piece of work. The digitalisation of a proportionate consenting process as well as the interview itself was imperative to this given the logistical impracticalities of face-to-face interviews. Utilisation of the Framework Method led to the aims and objectives of this study being met. It allowed theory and PPI input to shape the interview topic guides. This iterative approach was also used to develop the analytic framework and coding strategies within themes and sub-themes. Trials of interviews to test its ability to capture the true experiences of the participants were important in the early phases of the study, and insight from experienced research team members were invaluable to this. The PPI has been strong throughout the project but also in the development of this qualitative study. Meetings with the study PAG to finalise study documents maximised participation. Interview topic guides were reviewed at every stage to ensure that questions are easy to understand and yet, still address the aims of the study.

3.12.3 Considering the reflexivity of the researcher

It is necessary to consider the positional reflexivity of the researcher. To do this is to consider the experience of the researcher in terms of beliefs, values and ultimately, the influence on the research [183]. For context in this particular study, it was important to understand the researcher's role as a person in the interviews and accept that they are an active part of every step of the research [184].

Conducting an interview study utilised the researcher's strengths as a children's physiotherapist. One-on-one conversations with children were used to gain information regarding their condition and experience, this is common daily practice in the role of a children's physiotherapist. There are risks associated with the researcher being a health professional, with the main risk being that the clinical responsibility may 'overtake' the researcher. Discussing clinical issues with participants may feel like a way to engage conversation, but without guidance to stay on topic, would have led to an unsuccessful interview; for example, there was a risk it could have become more of a clinical consultation and overly directed. To manage this, regular discussions and reviews of interviews took place with supervisory team

members (SR, SP – named authors on the published paper). Both have a wealth of experience in conducting qualitative research, one of whom (SP) has extensive experience of qualitative research with children.

Reflexive logs were kept from the beginning of data collection. These logs allowed the researcher to review interviews in terms of how they felt and whether there was acceptable/suitable conversation throughout to promote information sharing. Logs were often based on discussions with supervisors as well as personal reflections and resulted in alterations in interview techniques. Interview skills, as well as reflections on the skills developed as data collection progressed. Most importantly to a point that would be beneficial in later studies within the programme of work. The final study includes a nested qualitative study within the feasibility study. The reflections included techniques such as altering questions to be more open-ended, and increased comfort with silences and pausing after questions, to allow participants the time to consider their thoughts and share what they saw fit.

3.13 Conclusion

The research set out to investigate the experiences and understanding of children/families and clinicians when considering non-surgical treatment of Perthes' Disease. Also, it aimed to explore the potential for a digital intervention to support self-management. Qualitative interviews were used to explore current care, the experiences of key stakeholders, and investigate the perceptions of key stakeholders when considering a digital self-management intervention.

Participants explained their exposure to the magnitude of the variation of care within this patient population. They discussed the impact that this variation of care had on them. Similarly, children and family participants highlighted positive impact that being involved in decision making had on their experience.

Children with Perthes' Disease, their families who care for them, and the clinicians who treat them, have all demonstrated a desire for more information to guide the non-surgical treatment. Most importantly, they have provided a positive response in

the data which informed the interim logic model in the development of a NON-STOP digital self-management intervention (Appendix I).

Chapter 4 – Clinical consensus recommendations for the non-surgical treatment of Perthes' Disease

4.1 Introduction

This chapter reports a modified Delphi study which produced clinical consensus recommendations for the non-surgical treatment of Perthes' Disease. The findings have been published as:

> Galloway, A. M., Keene, D. J., Anderson, A., Holton, C., Redmond, A. C., Siddle, H. J., Richards, S., & Perry, D. C. (2024). Clinical consensus recommendations for the non-surgical treatment of children with Perthes' Disease in the UK. *The Bone & Joint Journal*, *106-B*(5), 501-507. https://doi.org/10.1302/0301-620x.106b5.Bjj-2023-1283.R1

In reference to the MRC framework, this study maps to the "Developing the intervention" stage of the framework (shown in Figure 2.5). The Core elements of relevance at this stage were refine programme theory, after having updated the logic model (Appendix I), engaging stakeholders, included as participants and aiming to identify key uncertainties.

4.1.1 Background

The design, development and testing of a complex intervention includes multiple stages [44]. The approach used in this doctoral project has been presented in Chapter two. This chapter describes the second study. A modified Delphi study to develop clinical consensus recommendations for the non-surgical treatment of Perthes' Disease.

In Chapter three, the experiences of key stakeholders were explored, and demonstrated a range of topics displayed using themes. One of the themes included discussions around the variation of care for children with Perthes' Disease. Participants demonstrated their desire for agreement on what non-surgical treatment of Perthes' Disease should include. It is clear that there is a need for clinical

consensus on the non-surgical treatment of Perthes' Disease. Particularly when combined with the previous research highlighting the variation of care in the U.K. carried out by the researcher in preparation for this programme of work [28] and the priority setting by clinical speciality groups [32].

There is no robust evidence to guide clinicians on the optimal non-surgical treatment of Perthes' Disease [34]. In the absence of this, a Delphi study to achieve clinical consensus is an effective method. The results of this study form the majority of the 'clinical content' for the digital self-management intervention which is described later in this thesis (Chapter five). Whilst the results of this Delphi study inform the later stages of this programme of work, the lack of previous consensus makes the results of the study useful as a standalone piece of work. It will influence and support decision making for clinicians caring for children with Perthes' Disease. This has been supported by effective dissemination of results including with professional bodies and organisations in the children's orthopaedic clinical setting.

4.2 Aims and objectives

Aim

To develop clinical consensus regarding the Non-Surgical Treatment of Perthes' Disease.

Objective

a) Undertake a consensus study, using a modified Delphi Method, to identify best practice regarding the non-surgical treatment of Perthes' Disease.

4.3 Study design

This was a modified Delphi study, conducted to develop clinical consensus recommendations and points to consider for the non-surgical treatment of Perthes' Disease. The consensus methods used were consistent with those recommended and previously used in children's orthopaedics [136, 185-188]. The methods have been discussed in more detail in the methodology section in Chapter two.

Considering the epistemological stance within this study, it is well-aligned with the stance of the overall thesis, pragmatism [42], which is discussed in more detail in Chapter one. A pragmatic approach aligns with using a modified Delphi method to gather the views of clinical experts in the field where no robust evidence exists. This is useful for clinical guidance but also formed the content of the digital self-management intervention for non-surgical treatment of Perthes' Disease.

4.3.1 Rationale for the methodological approach

Clinical consensus does not exist for many orthopaedic children's conditions. There have been clinical consensus groups established within the British Society for Children's Orthopaedic Surgery (BSCOS) that have resulted in consensus statements being produced. Recommendations were produced for musculoskeletal infection [189], developmental dysplasia of the hip [190] and the assessment of the foot in children with cerebral palsy that used similar methods to this Delphi study [191]. There is an additional Delphi study that was completed in children's orthopaedics and published on the management of Congenital Talipes Equinovarus (CTEV) [187]. The Delphi study for CTEV focused on service delivery and interventions for the condition, not dissimilar to the aims of this study.

The Delphi study offers certain advantages that most techniques do not, arguably the most influential in the context of this Delphi study, is remote completion. Particularly given the sparsity and dispersion of clinical specialists treating relatively low numbers of Perthes' Disease in the UK. In the British Orthopaedic Surgery Surveillance (BOSS) study 63 centres in the UK treated 371 children with Perthes' Disease over a two-year

period [25]. Gaining insight from these centres remotely significantly increases the ease of completion when compared to methods such as consensus development conferences. For example, attempting to have clinical specialists in the same place, at the same time would pose major logistical implications. By contrast, the Delphi method allows these specialists to complete a survey from wherever they are. It therefore poses to increase ease of completion and potential response rate [137].

4.3.2 Survey design

The survey for this Delphi study was created in collaboration with a Survey Advisory Group (SAG). The SAG consisted of experts in this clinical area including specialist children's orthopaedic physiotherapists, nurses and surgeons. There was representation from across the UK (Edinburgh, Sheffield, Leeds, Liverpool, London and York. There was also input from an independent chair, a post-doctoral clinical academic physiotherapist, with experience of conducting Delphi studies using this methodology in the adult-orthopaedic population. The 'survey advisory group' used their expert opinion as well as the summary of evidence (Appendix J), to develop a series of 'domains'. The summary of evidence was created by the researcher using the most relevant literature available pertaining to the non-surgical treatment of Perthes' Disease. This included work completed by the researcher prior to the undertaking of this doctoral fellowship including a systematic review of the non-surgical treatment of Perthes' Disease [34], a case review to describe variation of care in the UK [28] and the preliminary findings from the qualitative interview study that had recently concluded at the time of creating the Delphi survey. The remaining literature included in the evidence briefing summary included the recently developed core outcome set for studies on Perthes' Disease [95], the BOSS study [25] and two papers considered to be key papers in the field of Perthes' Disease that compared surgical and non-surgical management [29, 30].

Several domains arose from the survey advisory group meetings, the domains that arose are displayed in Table 4.1 below. These domains included statements regarding the non-surgical treatment of Perthes' Disease for which participants would describe

their level of agreement/disagreement. The response-options ranged from 'strongly agree' to 'strongly disagree' on a five-point Likert-like scale. This method has been previously employed in Delphi studies within this patient population [187]. The survey advisory group met three times virtually to discuss domains and wording of the statements for the final survey. There was communication via email in between meetings for less time-consuming tasks such as document-reviews and wording clarification. One key decision that was made within these survey advisory group meetings was the definition of 'early' and 'late' stage for the purpose of this consensus study. The rationale for this was that advice given to families differs at different stages of the condition. The advice, particularly around weightbearing/high-impact activities such as trampolines and long-distance running, is something that is more relevant in the early stages of Perthes' Disease. In the early stages, such as necrosis and early fragmentation, the avascular bone is at high risk of microdamage [49, 192]. Any recommendations made around exercise/activity advice would therefore depend on the stage of the child's Perthes' Disease. With this in mind, a decision was made to classify 'early stage' as children with Perthes' Disease in the necrotic or fragmentation stage. Children were deemed 'late stage' if they were in the re-ossification or healed stage of the disease. This information was given to participants on the online platform used to host the survey to clarify for participants completing the survey what was meant by 'early' and 'late' stage Perthes' Disease for the purpose of this Delphi study.

Table 4.1 – Domains within clinical consensus study

Domain	**Topics covered**
Exercises	Strengthening exercises (early and late stage) Range of motion (ROM) exercises (early and late stage) Water-based exercises Functional ability exercises Who, when, where? (who/when/where to do exercises)
Physical activity	Recreational activities (early and late stage) Activity modification

Education/information sharing	Understanding of Perthes' Disease Pain management Weight management and nutrition Mental wellbeing
Input from other services	Referral to orthopaedics Referral to physiotherapy Multi-disciplinary team input Communication between children/families and clinicians School support
Monitoring assessments	ROM measurement Outcome measures Orthopaedic follow up

After the content for was finalised, the survey was uploaded to Online Surveys and formatted accordingly. Online Surveys has been used in Delphi studies previously and is provided by the University of Leeds [193]. The survey was then tested with a number of individuals (of differing experience of both the project and surveys) on a range of devices including internet browser on a computer, mobile phone and tablet. Testing in a number of ways allowed the researcher to check accessibility and the practical aspects of loading the survey. Minor corrections included re-sizing of logos and alterations to links to the aforementioned evidence briefing paper.

4.4 Sampling and recruitment

This modified Delphi study took place completely online and recruited a volunteer sample of children's-orthopaedic specialist clinicians.

The target population were UK-based clinicians (physiotherapists, consultant surgeons and clinical nurse-specialists) who worked in a clinical setting at least once a week where children with Perthes' Disease are seen. They also need to have at least two years of experience of treating children with Perthes' Disease. This timescale was based on recent incidence rates that will ensure they have had adequate exposure and therefore an understanding of usual care for this patient group [25]. The aim of

this study was to gain 'clinical' consensus; therefore children and their families were not eligible to complete the survey.

4.4.1 Eligibility criteria

Clinicians (UK-based physiotherapists, surgeons and clinical nurse-specialists) were eligible for inclusion if,

1) They worked in clinical setting at least once a week that manages children with Perthes' Disease.
2) They had at least two years' experience of treating children with Perthes' Disease.
3) They had access to relevant technology (digital device capable of loading and completing the online survey as well as email) to take part.

4.4.2 Recruitment

Participants were recruited using relevant special interest group mailing lists. BSCOS, which is open to all of the aforementioned clinical disciplines, as well as the Association of Paediatric Chartered Physiotherapists (APCP), a specialist group of children's physiotherapists practicing in the UK. A professional relationship exists with both of these specialist groups and permission from respective gatekeepers were sought as part of the study approval. An email was sent to mailing list members to briefly explain the study and instructions on how to take part. Social media was also utilised to maximise recruitment. Tweets tagging specialist interest groups were used and invited those interested to contact the researcher for more information regarding the study.

Purposive sampling was used in this Delphi study [173]. Using this sampling method maximised the heterogeneity of the sample and ensure optimal distribution of location and professional discipline was achieved whilst maintaining experience level. This method had practical benefits in that participants could be recruited using the methods outlined below without employing time-consuming tasks such as contacting

specific NHS centres for potential participants. There are limitations to this method, selection bias being the most likely given that participants are responding to an advert to take part [194].

4.4.3 Sample size

There are limited resources providing set guidance on the optimal number of participants for Delphi studies. An evidence synthesis in 2016 summarised the most commonly used methods for consensus and found that the most common sample size with Delphi studies was six to eleven participants [185]. The authors highlighted work that stated panels of eleven had inter-rater reliability (Kappa) indices of 0.76 [195], suggesting a high level of agreement [196]. Another study within the review by Wagonner et al reported that a panel with less than six members had limited reliability and a group of over twelve participants had an insignificant increase in reliability (no Kappa coefficient provided) [197]. Whilst the 'insignificance' is not quantified, it does suggest that a sample size or around six to eleven is sufficient, as demonstrated in a number of Delphi studies within the evidence synthesis. The authors of a study included in the evidence synthesis, Nair et al stated that Delphi panels must include an adequate number of participants and, whilst that can be hundreds, it should be at least ten [195].

For this study, the recruitment target was 12 to 15 surgeons/clinical nurse specialists and 12 to 15 physiotherapists. This was in an attempt to recruit 24-30 clinicians in total. This was deemed realistic, achievable and was likely to result in sufficient responses to meet the aims and objectives based on optimal Delphi study designs [185].

4.5 Consent

Informed consent was gained for all participants taking part in the study. Given the minimal risk to the participants, a proportionate consent process was adopted. This is in line with HRA guidance as well as methods employed in previously reviewed

studies within this fellowship [174]. The guidance in section 2.6 of the HRA document outlines that for self-completion survey studies proportionate consent may be demonstrated by the return of the survey itself. In this study, participants explicitly confirmed their agreement to take part, providing consent at a level above that deemed appropriately proportionate for this type of study. It was made clear to the participants that their involvement in this study would remain confidential and any findings shared will be anonymised. Participants followed the link outlined in the email/social media post that directed them to the survey. The first page of the survey included the participant information sheet followed by the consent page. To demonstrate informed consent, the participants ticked a box outlining their agreement to involvement in this study and will include a 'statement of agreement' which read as follows:

> *'I have studied the information provided in the participant information sheet and understand what will be required of 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 participation is voluntary, and I am free to withdraw from this study, unchallenged, at any time.'*

Confirming this statement acted as written consent for the participant, outlining explicitly that the participant agreed to their inclusion in the study including permission to use data collected in the way outlined in the participant information sheet.

4.6 Data collection

After the survey had been agreed, it included 87 statements mapping onto the five domains shown in Table 4.1: 'exercises', 'physical activity', 'education/information sharing', 'input from other services' and 'monitoring assessments for clinical practice'. Participants then responded to each statement using a five-point Likert-like rating scale, commonly used in this type of study [198, 199]. This is attached as Appendix K**.**

Using this scale, the participant indicated their agreement (or otherwise) with each statement using the following options:

1. Strongly agree
2. Agree
3. Neither agree or disagree
4. Disagree
5. Strongly disagree

Participants took part in two rounds of online data collection, and each round was open for three weeks. The survey was hosted using Online Surveys [193]**.** After completing the consent process, participants were presented with the short evidence briefing document (Appendix J). In the first round of data collection, the participants were asked to indicate their professional discipline (physiotherapist, nurse or surgeon). They then progressed to the online survey, which included a series of statements regarding non-surgical treatment approaches.

In the first round, there were free-text boxes in which participants were able to comment on the statements provided as well as their responses. Participants were able to suggest amendments or clarifications to the statement wording in preparation for round two. Any new suggested statements were discussed with the research team, and it was decided whether a statement would be included in round two. More detail on this process is in data analysis section 4.7.1.

Quantitative analysis for round one involved removing statements that achieved the pre-determined level of consensus, $\geq$75% for either 'agreement' or 'disagreement'. Agreement for this Delphi study was a response of 'agree' or 'strongly agree', similarly, 'disagreement' was a response of 'disagree' or 'strongly disagree'. A 'neither agree/disagree' option was provided.

For round two, a summary report (Appendix L) was sent to participants, providing a summary of the group's results from round one including the statements that achieved consensus. There was no display of specific scores for each of the statements either as a whole cohort or the individual's score for a particular

statement. The statements that did achieve consensus were removed from scoring in round two to minimise burden for participants. For round two there was no free-text box available due to the decision to not have a third round. The participants were asked to, once again, rate the statements using the five-point Likert-like scale as they did in round one. Participants were made aware of the new statements for round two, of which there were five. New statements were clearly marked with an asterisk on the online survey that outlined that this statement was new to this round of the Delphi based on round one free text response and review.

A consistent threshold of ≥75% for either 'agreement' or 'disagreement' was used for round two to establish consensus recommendations. In order to not lose the opportunity to report potentially meaningful information from this consensus study, 'points to consider' were also included. These were statements in which consensus was not achieved, yet the statement came particularly close and warranted some degree of reporting, The reason for including this is that the statements may not have passed the 75% threshold, but were close to doing so and had a large proportion of participants selecting 'neither agree/disagree' to that statement. Given the lack of robust evidence in this area, it was felt to be important to provide a list of these statements. This was in order to allow those utilising the data from this Delphi study to apply clinical discretion to the 'points to consider' as many may impact things like service provision in some areas, but not others.

There have been previous studies that have employed various methods of defining 'points to consider' or statements that did not achieve consensus. Many have used different approaches of reporting these statements including ranking all of the statements within the Delphi study in order of 'amount of agreement' [200]. Here, Anderson et al ranked the statements that did not pass their agreed threshold (70%) in order of the percentage of people that voted 'very important' and then 'important'. This is just one of many options in the available literature for Delphi studies, however a systematic review of [201] and a paper assessing the methodological appropriateness the Delphi design [136] reported that many thresholds used within the studies are somewhat arbitrary. An appropriate threshold to define 'points to consider' for this Delphi study in Perthes' Disease was discussed with members of the

survey advisory group and supervisory team. It was agreed that it was suitable to apply a similar, lower threshold used in other Delphi studies for consensus and use this as the criteria for 'points to consider' [202]. This was, therefore, defined as any statement that had 70% or more in either agree/disagree were classified as 'points to consider', of which there were three, these are clearly labelled in the tables within the results section.

An initial email reminder was sent if no response is received after one week of non-completion, and a second after two weeks. There was a period of three weeks between round one and two to allow for response analysis and any amendments to online surveys. A summary of the process can be seen in the flow chart below.

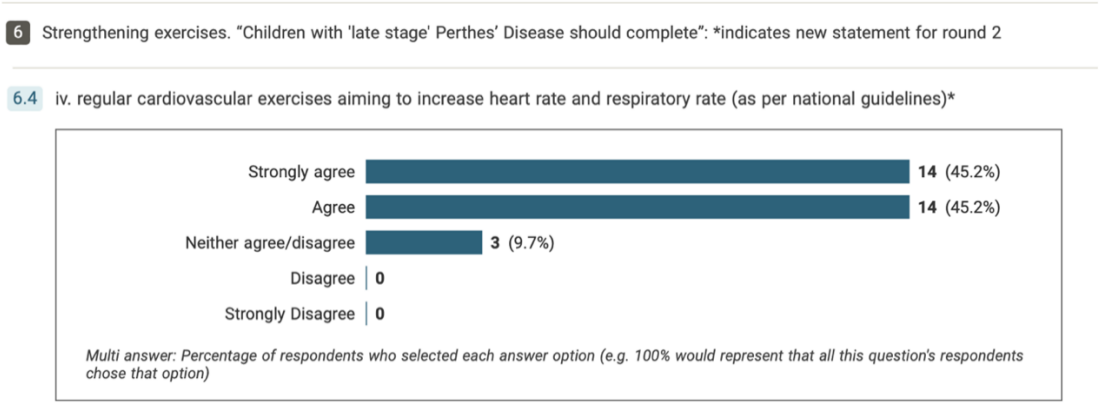
4.7 Data analysis

In this section, the method of analysis used for rounds one and two of this modified Delphi study are described.

4.7.1 Round 1

Free-text responses were analysed using content analysis [194]. This method involved the researcher reviewing the responses and identifying the presence of content that could be associated with one of the domains within the round one survey. In the event of a free-text response being something that did not align with a domain, this was to be discussed with the supervisory team and if appropriate, provided in a separate 'other' section in round two. Consistent with the methods described in other Delphi studies [203], these statements were organised in a document in sections depending on the domain they described. Where responses were relevant, they were used to form a new statement for the second round of the study. An example is shown in Figure 4.1 below:

Figure 4.1 – Example of survey with new statement for round two



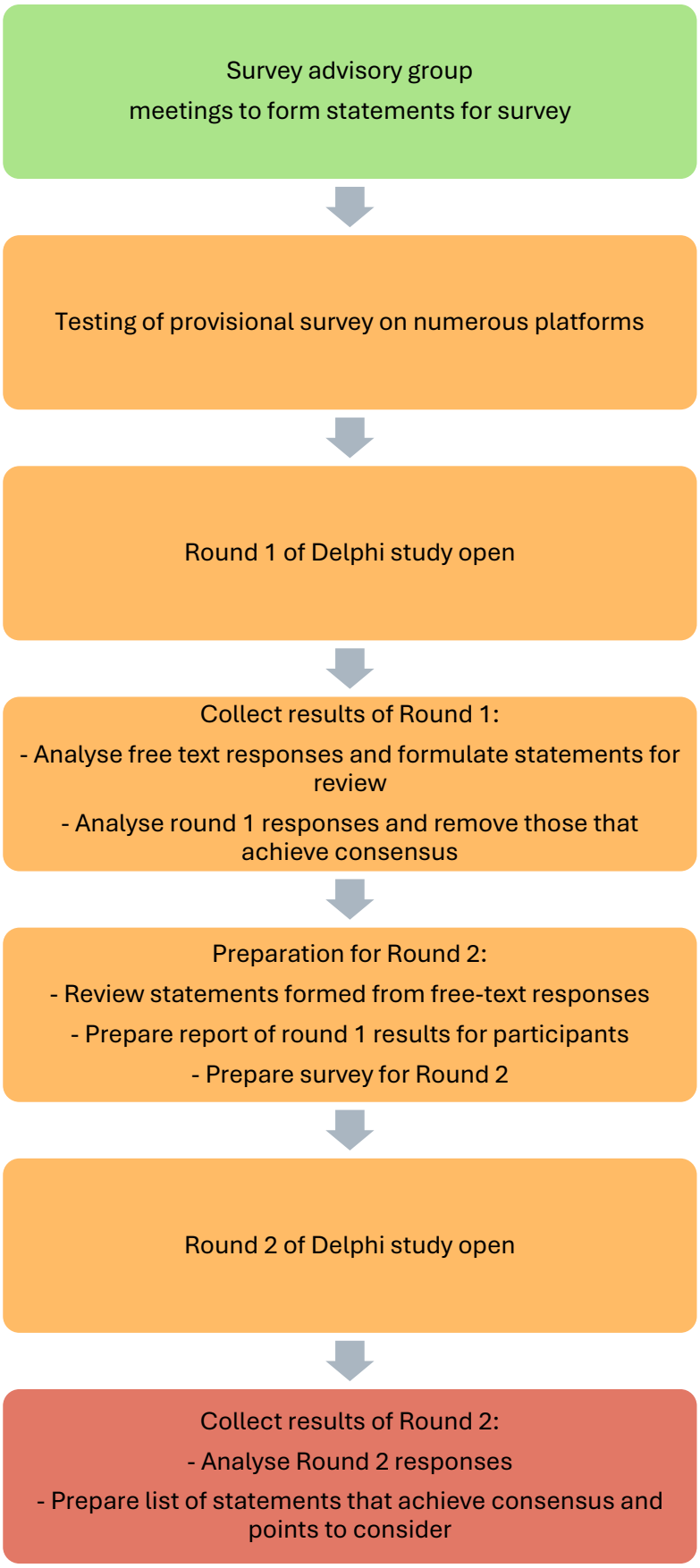
In this example, a free-text response suggested that children should complete exercise that increases respiratory rate and heart rate. This was analysed by the researcher and checked with members of the supervisory team, and deemed appropriate to include in round two and achieved consensus.

4.7.2 Round 2

Round two analysis did not include any qualitative aspects due to the removal of the free-text boxes. The quantitative analysis method was the same as in round one, with statements achieving clinical consensus presented within the domain in which they sat. The only addition in round two was that the final list of statements that were between 70-75% consensus were deemed 'points to consider' in the final list of statements.

An overview of the process followed within this Delphi study is shown below in Figure 4.2

Figure 4.2 – Flow chart showing process of Delphi



4.8 Ethics

This study recruited from professional bodies (BSCOS and APCP) and not through NHS services. It, therefore, required approval from the University of Leeds [204]. This was granted on 25/10/2022 and assigned the reference MREC 22-003.

4.9 Results

In round one, 40 participants responded to the survey. Nine did not respond to round two. More information on participants for each round can be seen in the table below.

Table 4.2 – Participant characteristics for rounds one and two

	Round 1	**Round 2**
Profession	**Number of participants (%) n = 40**	**Number of participants (%) n = 31**
Physiotherapist	22 (55)	19 (61)
Orthopaedic surgeon	17 (43)	11 (36)
Clinical Nurse Specialist	1 (2)	1 (3)

4.9.1 Round 1

After 40 participants responded in round one, there was clinical consensus in 31 statements across the five domains which were excluded from round two. There were nine statements in 'exercises' that achieved consensus; five statements in 'physical activity'; ten in 'education/information sharing'; five in 'input from other services' and two in 'monitoring assessments for clinical practice'. Sections below report the final list of consensus statements after completion of all rounds, with detail on the percentage of consensus achieved.

There were 38 free-text responses after round one. From these, six were presented to the supervisory team for discussion on inclusion in round two. Three of these statements were included in round two as new statements. One in the domain 'exercises' and two in the domain 'Education/information'. The statement in the

'Exercises' domain was appropriate to include in both the 'early stages' and 'late stages' of Perthes' Disease, creating a fourth additional statement.

The reasons that were excluded included statements outlining current clinical practice which was not relevant to this Delphi study or were outside of the scope of this Delphi study. An example of this was discussion around exclusion of other conditions.

It was made clear to participants that the statements were new to round two at the point of responding. There was also information in the report following round one (Appendix L) to remind participants that there were new statements for round two.

4.9.2 Round 2

31 participants responded to 60 statements in round 2 of the Delphi study (see Table 4.1). After round two, there 45/91 statements that achieved consensus within the five domains. This section will outline the statements that achieved clinical consensus and are recommendations for clinical practice.

4.9.2.1 Exercises

14/32 statements in the 'Exercises' domain achieved consensus, and one point to consider, as shown in table 4.3 below.

Table 4.3 – Consensus statements in the 'Exercises' domain

Exercises		
Statement	**Outcome**	**%**
a. Strengthening exercises. "Children with 'early stage' Perthes' Disease should complete":		
i) hip strengthening exercises	No consensus	
ii) knee strengthening exercises	No consensus	
iii) foot & ankle strengthening exercises	No consensus	
iv) trunk strengthening exercises	No consensus	
v) high impact strengthening exercises (e.g. squat-jumps, star-jumps)	Disagree	83

vi) any strengthening exercises as long as they avoid discomfort	No consensus	
vii) regular cardiovascular exercises aiming to increase heart rate and respiratory rate (as per national guidelines)	Agree	78.1
b. Strengthening exercises. "Children with 'late stage' Perthes' Disease should complete":		
i) hip strengthening exercises	Agree	85.4
ii) knee strengthening exercises	Point to consider	70.7 agree
iii) foot & ankle strengthening exercises	No consensus	
iv) trunk strengthening exercises	Agree	78.1
v) high impact strengthening exercises (e.g. squat-jumps, star-jumps)	No consensus	
vi) any strengthening exercises as long as they avoid discomfort	No consensus	
vii) regular cardiovascular exercises aiming to increase heart rate and respiratory rate (as per national guidelines)	Agree	87.5
c. Range of movement stretching exercises. "Children with 'early stage' Perthes' Disease should complete":		
i) hip stretches	Agree	90.3
ii) knee stretches	No consensus	
iii) foot and ankle stretches	No consensus	
iv) spinal stretches	No consensus	
v) any stretching exercises as long as they avoid discomfort	No consensus	
d. Range of movement stretching exercises. "Children with 'late stage' Perthes' Disease should complete":		
i) hip stretches	Agree	78.1
ii) knee stretches	No consensus	
iii) foot and ankle stretches	No consensus	
iv) spinal stretches	No consensus	
v) any stretching exercises as long as they avoid discomfort	Agree	75.1
e. Water-based exercise. Children with Perthes' Disease should be advised to complete		
i) supervised (physiotherapy-led) water-based exercise	No consensus	

ii) water-based exercise as self-management, i.e., prescribed exercises in a local pool (not supervised by a physiotherapist)	Agree	82.9
iii) water-based exercise when land-based physiotherapy is not effective	Agree	80.5
f. Functional ability exercises		
i) Children with Perthes' Disease should complete balance exercises	Agree	84.4
ii) Children with Perthes' Disease should receive gait education	Agree	84.4
iii) Children with Perthes' Disease should have advice on potential use of mobility aids	Agree	82.9
e. Who, when, where? Children with Perthes' Disease should complete prescribed exercise regimes		
i) at home under the supervision of parent/family members	Agree	85.4
ii) at school under the supervision of school-staff members	No consensus	
iii) at the hospital under the supervision of clinicians	No consensus	
iv) at extracurricular activities (e.g., sports clubs, etc.) under the supervision of those leading the activities	No consensus	

4.9.2.2 Physical activity

9/20 statements in the 'Physical activity' domain achieved consensus, as shown in table 4.3 below.

Table 4.4 – Consensus statements in the 'Physical activity' domain

Physical activity		
Statement	**Outcome**	**%**
a. Recreational activities. "In the early stages of Perthes' Disease, the following activities should be discouraged":		
i) Swimming	Disagree	78
ii) Contact sports (e.g. football, rugby)	Agree	78.2
iii) Other team sports (e.g. basketball, netball, hockey)	No consensus	
iv) Long distance running (more than 1-2 mile)	Agree	75.6
v) Horse riding	No consensus	
vi) Cycling	Disagree	84.4
vii) Gymnastics	No consensus	

viii) PE at school	No consensus	
ix) High-impact activities (e.g. bouncy castles and trampolines)	Agree	90.6
b. Recreational activities. "In the late stages of Perthes' Disease, the following activities should be discouraged":		
i) Swimming	Disagree	80.6
ii) Contact sports (e.g. football, rugby)	No consensus	
iii) Other team sports (e.g. basketball, netball, hockey)	No consensus	
iv) Long distance running (more than 1-2 mile)	No consensus	
v) Horse riding	Disagree	75.1
vi) Cycling	Disagree	80.4
vii) Gymnastics	No consensus	
viii) PE at school	No consensus	
ix) High-impact activities (e.g. bouncy castles and trampolines)	No consensus	
b. Activity modification." Children with Perthes' Disease should":		
i) use a walking aid (e.g. crutches, Zimmer—frame) to modify their activity if symptoms (pain, limping, reduced activity) persist	Agree	80.5
ii) use a wheelchair to modify their activity if symptoms (pain, limping, reduced activity) persist	No consensus	

4.9.2.3 Education/information sharing

13/20 statements in the 'Education/information sharing' domain achieved consensus, and one point to consider, as shown in table 4.4 below.

Table 4.5 – Consensus statements in the 'Education/information sharing' domain

Education/information sharing		
Statement	**Outcome**	**%**
a. Understanding of Perthes' Disease. "Clinicians should provide children/families with":		
i) information regarding the disease process including the affected anatomical structures and prognosis	Agree	100
ii) information regarding current research relating to Perthes' Disease including aetiology and epidemiology	Agree	85.3

iii) information regarding where additional patient and family information resources can be found (e.g., STEPS website)	Agree	100
b. Pain management. "Children with Perthes' Disease should be":		
i) advised to take paracetamol or equivalent for pain management	Agree	92.7
ii) advised to take ibuprofen or equivalent for pain management	Agree	81.7
iii) advised to take morphine or equivalent for pain management	Disagree	75.6
iv) advised on pacing and activity levels	Agree	97.6
v) advised on the use of heat/cold therapy	Agree	75.6
vi) advised on the use of TENS/equivalent electrotherapy	No consensus	
vii) encouraged to use massage to aid pain relief	No consensus	
viii) prescribed steroid injection for pain management	No consensus	
ix) be referred to a pain management service if their symptoms are not managed with medication and/or physiotherapy	Point to consider	71.9 agree
x) be provided with resources on chronic pain for persistent pain related to Perthes' Disease (where general Perthes' advice is not relevant/effective)	Agree	87.6
c. Weight management and nutrition		
i) Children with Perthes' Disease should receive advice on lifestyle, weight management and nutrition from a healthcare professional	Agree	75
ii) Children with Perthes' Disease should receive advice on lifestyle, weight management and nutrition only when indicated	No consensus	
iii) Children with Perthes' Disease should be referred to a specialist service for weight management and nutrition when clinically indicated	Agree	87.8
iv) Monitoring weight management and nutrition is an essential part of treatment i.e. height and weight at all appointments	No consensus	
v) Information from reviews such as activity-related information, weight management advice or wellbeing support, should be shared with school	No consensus	
d. Mental wellbeing. "Children with Perthes' Disease and their families should be":		

i)	given the opportunity to discuss their (or their child's) mental wellbeing with any healthcare profession	Agree	95.1
ii)	signposted to general mental wellbeing resources (e.g., the STEPS charity website or NHS 111 website)	Agree	92.7

4.9.2.4 Input from other services

7/16 statements in the 'Education/information sharing' domain achieved consensus, as shown in table 4.5 below.

Table 4.6 – Consensus statements in the 'Input from other services' domain

Input from other services			
Statement		**Outcome**	**%**
a. Referral to orthopaedics			
i)	Any child with suspected Perthes' Disease should be referred for specialist review	Agree	97.6
ii)	Any child who does not improve from a symptom/symptom-management perspective should have access to an orthopaedic specialist	Agree	97.6
iii)	Children with Perthes' Disease should be centralised to a team with a specialist interest in Perthes' Disease in their geographical region	No consensus	
b. Referral to physiotherapy. "Children with Perthes' Disease should":			
i)	be offered an initial assessment with a physiotherapist	Agree	97.6
ii)	be seen by a physiotherapist regularly until the disease process is complete/healing is observed	No consensus	
iii)	keep regular activity diaries to share with healthcare professionals	No consensus	
iv)	be seen by a physiotherapist until they can self-manage independently	Agree	75
c. Multi-disciplinary team input. "Children with Perthes' Disease should":			
i)	be offered an assessment from an occupational therapist	No consensus	
ii)	be offered an assessment from a social worker	No consensus	
iii)	be offered an assessment from a psychologist	No consensus	
iv)	have a named clinical 'key worker' regardless of MDT role	No consensus	

d. Communication between patients and clinicians		
i) Children with Perthes' Disease and their families should have a means of direct communication with clinicians between appointments	Agree	80.5
ii) Children with Perthes' Disease and their families should be directed towards means of contacting other children with Perthes' Disease and their families i.e. peer-support groups/forums	Agree	95.1
e. School support. "Children with Perthes' Disease should have access to a":		
i) physiotherapist in a school setting	No consensus	
ii) nurse in a school setting	No consensus	
iii) named school-support staff member	Agree	81.2

4.9.2.5 Monitoring assessments for clinical practice

2/3 statements in the 'Education/information sharing' domain achieved consensus, and one point to consider, as shown in table 4.6 below.

Table 4.7 – Consensus statements in the 'Monitoring assessments for clinical practice' domain

Monitoring assessments for clinical practice		
Statement	**Outcome**	**%**
a. Clinical assessments. "Children with Perthes' Disease should have":		
i) their ROM documented at every appointment (regardless of MDT role)	Agree	78
ii) a validated quality of life assessment tool completed at initial assessment and regular intervals	Agree	83
iii) regular reviews with an orthopaedic specialist until they reach skeletal maturity	Point to consider	73.2 agree

4.9.3 Differences between professions

In both round one and two, there was a single clinical nurse specialist (CNS). Otherwise, in round one the cohort was divided into 22 physiotherapists and 18 surgeons. In round two there were 19 physiotherapists and 12 surgeons. Stratifying the results by profession did not make for many differences in outcome. Below are two tables that demonstrate the statements in which there was consensus when

stratifying by profession. Table 4.7 shows the statements that achieved consensus when looking at the level of agreement/disagreement amongst the physiotherapists. Table 4.8 shows the statements that achieved consensus when assessing the level of agreement/disagreement amongst surgeons and CNS.

Table 4.8 – Statements of consensus amongst physiotherapists

Statement	**Original outcome**	**Outcome after stratifying**
Children with 'early stage' Perthes' Disease should complete hip strengthening exercises	No consensus	Consensus agree (77.4%)
Children with 'late stage' Perthes' Disease should complete any strengthening exercises as long as they avoid discomfort	No consensus	Consensus agree (79%)
Children with Perthes' Disease should be referred to a pain management service if their symptoms are not managed with medication and/or physiotherapy	Point to consider (71.9%)	Consensus agree (84.2%)

Table 4.9 – Statements of consensus amongst surgeons and CNS

Statement	**Original outcome**	**Outcome after stratifying**
Children with 'late stage' Perthes' Disease should complete knee strengthening exercises	No consensus	Consensus agree (76.9%)
Children with 'late stage' Perthes' Disease should complete knee stretching exercises	No consensus	Consensus agree (76.9%)
Children with Perthes' Disease should be prescribed a steroid injection for pain management	No consensus	Consensus disagree (76.9%)
Children with Perthes' Disease should have regular reviews with an orthopaedic specialist until they reach skeletal maturity	No consensus	Consensus agree (84.6%)

4.10 Discussion

Variation in care negatively impacts children with Perthes' Disease and their families; this was a significant theme in the recently published qualitative study [155]. Family members and clinicians expressed their desire for consensus after outlining a sense of disagreement in the community. In response, this Delphi study was completed. The

aim was to develop clinical consensus recommendations with the aim of reducing the variation of care and provide insight as to what best practice may involve based on the opinion of experts. The consensus study helped to define the key intervention functions for a self-management intervention by identifying priority areas of clinical practice. These were then ready to integrate into the digital self-management intervention. They include stretching and strengthening exercises for the affected hip and education regarding pain management, wellbeing, and disease progress.

4.10.1 Exercise recommendations

Exercises for lower limb strengthening and stretching exercises gained clinical consensus. Clinicians agreed children with 'early' and 'late' stage Perthes' Disease should complete hip stretches. In 2006, Brech and Guarnieiro evaluated the effects of physiotherapy for children with Perthes' Disease and concluded that children that received a stretching programme had an increased range of joint motion (ROM) [156]. The study from 2006 does have limitations, with a small sample size in the treatment group (n=17) and no clear guidelines on intervention content other than the fact that participants attended twice weekly physiotherapy sessions. There was also no comment on the stage of the disease of participants. That being said, the participants that completed a stretching programme had increased ROM at follow up. Which is positive for providing non-surgical containment of the femoral head given the need for decent amounts of abduction and rotation at the hip to achieve containment [205]. Similarly those that completed a lower limb strengthening programme in the Brech and Guarnieiro study demonstrated an increase in muscle strength. Particularly when assessing the strength in rotational movements of the hip. These muscles are, important in non-surgical containment of the hip joint. Having strength in muscles around the hip can reduce articular dysfunction [156]. Studies in similar conditions such as femoroacetabular impingement syndrome have demonstrated that maintaining strength in muscles that abduct the hip allow it to move within the available range [206]. In relation to Perthes' Disease, it is plausible to suggest that maintaining strength could lead to using the available range and reduce the risk of muscle shortening that comes with changes to the femoral head.

4.10.2 Activity recommendations

There was clinical consensus for reducing high-impact activities such as contact sports, trampolines and bouncy castles, and long-distance running (> 1 mile) in the 'early stages' of Perthes' Disease. There are no studies that look at activity modification specifically, however many clinicians incorporate it into their non-surgical treatment in clinical practice [207]. With the rationale being that a soft femoral epiphysis combined with increased loads can lead to femoral head deformity [208].

Hydrotherapy is a treatment option available to some physiotherapists, and has been found to be successful when used in some health-conditions for children. In a study examining the effect of hydrotherapy on children with Autistic Spectrum Disorder (ASD), there were significant improvements in behaviours affecting mental health and wellbeing following a period of hydrotherapy [209]. In 2005, a randomised controlled trial explored the effectiveness of hydrotherapy for children with Juvenile Idiopathic Arthritis. The authors demonstrated improvements in quality of life scores and cardiovascular fitness for those receiving hydrotherapy [210]. It significantly reduces the forces through the hip and knee, with some data showing joint forces are reduced by up to 55% in dynamic exercises such as jumping in water [211].

As discussed above, relieving the loading forces through the hip joint are widely accepted as an important aspect of treatment in the early phase of Perthes' Disease. This is irrespective of whether a surgical or non-surgical approach is used [212]. Combine the benefits of water-based activity that exist in the literature, with the consensus in this Delphi to reduce high-impact activities and sports, then it is not surprising to have achieved clinical consensus for the use of water-based activities for self-management and swimming. All of the findings discussed do lead to questions about the implementation of water-based activities for children with Perthes' Disease. Specifically, does this need to be formalised hydrotherapy, or exercises prescribed to complete in water. Alternatively, and perhaps more pragmatically when considering the burden to healthcare services and families, general advice from

healthcare professionals to children with Perthes' Disease and their families about regular swimming. Future research could evaluate the effectiveness and differences in experience of those carrying out therapist-supervised hydrotherapy for Perthes' Disease when compared with a general recreational swimming programme.

4.10.3 Education/information sharing

In Chapter three, which describes a qualitative study to understand key stakeholders' experience of care for Perthes' Disease, there was a recurrent theme which reported the need for consistent messaging, and information for children with Perthes' Disease and their families. Participants recounted the experience of different clinicians giving different advice to patients. In this Delphi study, 13 of the 45 statements that achieved clinical consensus fell within the 'Education/information sharing' domain. The over-arching programme of work in this project is to develop a self-management intervention that will include a significant amount of education and advice for children and their families. The content of this will be derived with a focus on this Delphi study's findings.

4.10.4 Input from services

This Delphi study achieved clinical consensus and recommended that all children with Perthes' Disease should receive at least an initial assessment with a physiotherapist. There was also consensus that the child should be seen by a physiotherapist until they can self-manage independently. These findings contrasted with the variation in care in the UK for Perthes' Disease identified in a previous case review study by the researcher [28], in which almost 20% of participants were not referred to a physiotherapist. With effective dissemination of this Delphi study's findings amongst specialist interest groups in the UK, there is a potential to reduce some of the variation of care for these children. By providing recommendations to clinical specialists based on this consensus study, more children with Perthes' Disease could have physiotherapy input. Other than the study discussed previously from Brech and Guarnieiro, there are no robust studies demonstrating which types of physiotherapy

input are most effective, as demonstrated in the systematic review published by the researcher [34]. Future research could compare optimised non-surgical treatment of Perthes' Disease with care provision nationally and internationally.

As discussed, the prior qualitative study demonstrated the high degree of importance families put on education related to the condition and being involved in their care. Education concerning the disease progress was something clinicians agreed is paramount in the treatment of Perthes' Disease, but this relies on access to specialist services. There was a strong consensus (>90% of clinicians) towards support for mental wellbeing of a child with Perthes' Disease. Access to mental health support is challenging for children and young people in the UK. A study by Hines et al. demonstrated that in 2019 only 46 (26.6%) of acute hospitals had access to paediatric liaison psychiatry services [213]. Whilst this may have improved since, it certainly stands to reason that alternative approaches for delivering mental health support are needed. These could include signposting to relevant third sector organisations and online support. In addition, clinicians could potentially include mental wellbeing advice within the self-management support they provide. The qualitative study in Chapter three highlighted the thoughts and experiences of key stakeholders, involvement in decision making and feeling as though they are a part of their own care was important. Future research could re-visit the thoughts of children with Perthes' Disease, their families, and the clinicians who care for them after optimising this information provision for things such as mental wellbeing and available support.

4.10.5 Monitoring assessments

Clinicians who took part in this Delphi study agreed that children with Perthes' Disease should have their hip ROM assessed at every appointment. It is understood that maintaining range of motion at the hip joint allows for better joint congruency due to the impact of the immature femoral head against the acetabulum [64]. Being able to identify when ROM changes is of importance to clinicians, a reduction in ROM could indicate worsening joint deformation [214]. In this situation, a deformed femoral head can cause altered ROM where anatomical structures come into contact

and rather than the femoral head and acetabulum acting as a 'ball' and 'socket', the femoral head 'hinges' off of the acetabulum. Often referred to as a complex form of femoroacetabular impingement [215]. This can cause significant pain for the child, and also direct treatment options. Increased femoral head deformity is a key criteria when considering whether a child with Perthes' Disease should have surgery [216]. This is likely to explain why regular review of ROM gained clinical consensus in this Delphi study.

4.10.6 Differences in professional groups

The number of participants in this Delphi study allowed some more in-depth analysis within sub-groups. As seen in section 4.9.3, it was possible to stratify the results of the Delphi study by profession. This was separated into physiotherapy participants and remaining participants. For the purpose of data analysis, the single CNS' results were analysed with surgeon responses.

There were not many differences in terms of what achieved or did not achieve clinical consensus when stratifying for professional background. In the physiotherapy group, seen in Table 4.8, there were three statements that achieved consensus, all were for agreement. Two statements relate to the prescription of strengthening exercises. There is good reason to believe this achieved consensus for agreement due to the nature of the physiotherapy role. Provision of exercises for strengthening is very common practice, with literature supporting the use of strengthening exercises for children with Perthes' Disease [156]. The final statement that achieved consensus related to onward referral to pain management services for children with Perthes' Disease who have persistent symptoms. Similar to the exercise statements, the whole cohort responses were not skewed towards disagreement, rather they had a higher proportion of 'neither agree or disagree' which meant the statement did not reach the consensus threshold. This could be due to the understanding of services like pain management from other members of the MDT. For instance, very few physiotherapists see solely orthopaedic complaints and often refer into services like this. Whereas in the orthopaedic surgery setting, the caseload is very specialised and

whilst no robust data exists, anecdotally it is reasonable to suggest their exposure to this service is less. Research to explore the understanding of MDT services as well as availability of services would be needed to understand the true reasons for this.

Table 4.9 outlines the differences when stratifying for remaining participants. This table has two statements pertaining to knee stretches/strengthening in 'late stage' Perthes' Disease that would have reached consensus without physiotherapist involvement. It is not clear why the physiotherapy sub-group would have altered the consensus level for this statement. Future research could explore this further. However, in table 4.7, the statement 'any strengthening exercise as long as they avoid discomfort' achieved consensus. It is possible to suggest that the statement would address these knee exercises in 'late stage' Perthes' Disease.

In a similar way to the physiotherapy statements largely being role-specific statements, such as exercises and other closely linked services like referral to occupational therapy, there is a slight pattern to the remaining participants' sub-group. Statements around medicines use (steroid injection) and reviews within orthopaedic clinics (often surgeon/CNS-led) achieved consensus when removing physiotherapy participants. It is also worth noting there were other statements around medication that achieved consensus in round one when stratifying for remaining participants. These did however achieve consensus in round two. It is possible that this too relates to role specific duties. Physiotherapists rarely prescribe or recommend the use of medication because it is not a part of typical training, rather a specialist training that a small portion of specialists receive in postgraduate settings. Physiotherapists also receive additional training on things such as pain management, pacing and other holistic approaches to pain management. Their expertise in this area are slightly wider-reaching than medications and so could explain why their views on medication are somewhat neutral.

4.10.7 Strengths and limitations

For the first time, a clinical consensus study has taken place for the non-surgical treatment of Perthes' Disease. Previous work as part of this programme of research

(see Chapter three) has highlighted the desire from clinicians and key stakeholders for clinical consensus. Through robust methods, utilising innovative online recruitment and completion, this Delphi study has achieved clinical consensus in a number of domains. Using professional specialist societies and social media to recruit, it led to a diverse sample of clinicians from the breadth of the UK and achieved a total number of participants above the target of 24-30 participants (40 participants in round one, 31 in round two). Whilst this number is not in the same region of other consensus studies for children's orthopaedic conditions [187], Perthes' Disease is a much less commonly seen condition. Nevertheless, this study achieved a sample size that is recommended in the literature for the optimum number of participants for a Delphi study [136]. Another strength of this Delphi study in comparison to other children's orthopaedic consensus studies is that it recruited a relatively equal number of physiotherapists compared to orthopaedic surgeons. This maximised the heterogeneity of the sample and in turn how effectively the results of this Delphi represent the population that care for children with Perthes' Disease.

The sampling method used in this Delphi study is a potential limitation. Two methods of non-probability sampling could have been used to minimise bias. In this Delphi study, there was a naturally occurring mitigation for this risk. Snowball sampling happened as a result of the advert for the study being shared on social media (as per the protocol) and some clinical specialists contacted the researcher to explain that they had shared the invite with their clinical team [217]. It was not possible to track these participants to monitor where they were recruited from. Nevertheless, all participants were vetted in the same way and confirmed adequate level of experience and provided demographic information to optimise heterogeneity. Self-selection was a limitation that the researcher and supervisory team was aware of and accepted. Mostly because the nature of this study was to recruit specialists treating Perthes' Disease, a condition that is not treated by every children's-orthopaedic specialist in the U.K.

Divergence between sub-groups is a limitation of this particular Delphi study. The particular risk in this Delphi was that with a mix of professions, there would be a difference of opinion in either direction that would result in a lack of consensus. To

mitigate for this, sub-analysis was completed with sub-groups based on professional background. Whilst there are not many, there are differences in consensus when stratifying by professional background as shown in section 4.9.3. An additional risk in completing this sub-analysis is the of this nature is difficult to mitigate for entirely. The key way that this Delphi study mitigated for this, was to ensure that there were adequate numbers of each sub-group to deliver consensus statements that could be representative of the population (children's orthopaedic specialists). With that in mind, samples were based on the literature available in previous children's orthopaedic Delphi studies [187, 189]. Other methods used in consensus studies such as consensus development conference or nominal group technique have the potential to promote discussion and resolve areas of uncertainty [129]. This comes with the inherent risk of increased bias due to over-represented participants willing to share their views more than others. Whilst the divergence is accepted as a limitation, the sample size achieved within the sub-groups was adequate enough to produce consensus recommendations as a whole cohort, and in sub-group analysis. The importance of this is primarily to the professional backgrounds represented. For instance, physiotherapists may be interested to know that the viewpoint of colleagues in this field is that children with 'early stage' Perthes' Disease should complete any strengthening exercises as long as they avoid discomfort.

Two rounds of the Delphi were used; a third round of Delphi could have been useful to try and achieve consensus in some of the statements that got close to 75%. It is also fair to say that the risk of losing participants to follow up would also have increased, as has been shown in many previous Delphi studies [218]. In a similar attempt to increase the number of statements that achieved clinical consensus in a modified Delphi study, the option of 'neither agree/disagree' could have been removed from the Likert-like scale. It is not required to always have this, and can be altered to suit the needs of the study [198, 199]. Giving participants an option to remain neutral is an important thing to do when there is a potential for participants to not have an opinion on a matter, or not take part in the activity as part of their practice. For example, if we were to ask physiotherapists about delivering a certain type of osteotomy, it would be important to have a neutral option such as 'not

applicable' or 'neither agree/disagree'. It is not common practice for physiotherapists to be involved in the surgical planning aspect of care for children with Perthes' Disease. In this Delphi study, it is reasonable to suggest that every participant is involved in the aspects of care that are explored in the statements and therefore could give an opinion.

4.10.8 Implications for practice and future research

This Delphi study was undertaken to inform the content for the digital self-management intervention. It is also an important standalone project that can be used to guide clinical practice as it is the first study to produce clinical consensus recommendations in the non-surgical treatment of Perthes' Disease. Since the completion of this study, the work has been published in a peer-reviewed orthopaedic journal [219]. The findings have also been shared via social media, at national conference and summarised as an infographic (Appendix M). This was done to provide clinical centres with the information from this Delphi study which could help design and plan clinical services as well as educate families. For example, departments may be able to consider their resources for information sharing with families based on the statements from this consensus study. Further research is needed to help implement these findings in clinical practice. Including reviewing the uptake and effectiveness of any changes in practice.

Moving forward clinical research should look to utilise the findings from this Delphi study in any non-surgical intervention involved in research in the UK. As demonstrated in a systematic review by the researcher [34], there is no robust evidence to support one particular non-surgical treatment method for Perthes' Disease. In the absence of robust evidence, the clinical consensus statements from this Delphi study should be considered and implemented in to a non-surgical intervention.

4.10.9 Considering the reflexivity of the researcher

As a children's physiotherapist, there is a potential that the survey itself could have been influenced by the researcher's professional background. This could have included influences such as the inclusion of exercises or activities that the researcher has a personal experience of and preference for treatment. The survey advisory group acted as the most significant mitigation for this. There were additional physiotherapists in the group for the design of the survey, which ensured an equal influence, as did the presence of other disciplines such as surgeons and clinical nurse specialists.

There was also a potential risk that the survey was purely focused on the development of a digital self-management intervention and not aligned with the aims and objectives of this Delphi study. The aim of this Delphi study was to develop clinical consensus recommendations for the non-surgical treatment of Perthes' Disease. A key role of the survey advisory group was to ensure that the survey met the aims and of objectives. Regular meetings and discussions with the supervisory team also assisted with this. Regular reviews of the Delphi study protocol were also completed.

4.11 Conclusion

The aim of this Delphi study was to produce clinical consensus recommendations on the non-surgical treatment of Perthes' Disease and points to consider. Forty-five statements in which clinical consensus was reached, and three points to consider, have been generated, and thus, the aim of this study was achieved.

The 45 statements were made up of 14 statements relating to exercises, nine relating to physical activity, 13 relating to education/information sharing, seven relating to input from other services and two relating to monitoring assessments. There is a known absence of robust evidence, and with that in mind, these recommendations are useful to guide clinical practice in the non-surgical treatment of Perthes' Disease. The findings have been disseminated accordingly amongst clinical experts in children's orthopaedic care [219].

The findings of this clinical consensus study informed the content for the development of the NON-STOP digital self-management intervention described in the next chapter. The findings from the qualitative study in the previous chapter and patient and public involvement activity optimised the development of a novel digital self-management intervention, ensuring it meets the needs of those using it.

Chapter 5 – Producing the digital self-management intervention: The NON-STOP app

5.1 Introduction

This chapter describes the design and development process of the digital self-management intervention for children with Perthes' Disease, delivered via a smart-device app, hereby referred to as the NON-STOP app. At the time of submission has been accepted for publication in August 2025:

> Galloway, A. M., Anderson, A., Casimir, E., Holton, C., Redmond, A. C., Keene, D. J., Redmond, A. C., Siddle, H. J, Richards, S., & Perry, D. C. (2024). From theory to practice: Insights into intervention development of the NON-STOP app for children with Perthes' Disease. *Bone & Joint Open.*

The theoretical underpinning of the overall programme of work was pivotal in this design stage of the NON-STOP app, as was a detailed understanding of the context in which children and their families would use this technology. Design and development work is underpinned by a programme theory, defined in section 5.3 [220] (in the form of an updated logic model), and enhanced by the introduction of behaviour change theory.

There are many approaches to intervention development [138]. This chapter begins by briefly describing some of them, with a primary focus on the approach used in this thesis. The development and design process included the creation of app content and training materials for app users and conducting preliminary user-testing of the prototype NON-STOP app. These steps were an integral part of app development by the software providers. This was done in preparation for the final study, a mixed-methods study to test the usability and acceptability of the NON-STOP app.

In reference to the MRC framework, this study maps to the "Developing the intervention" stage of the framework (shown in Figure 2.5). The core elements of relevance at this stage were engaging stakeholders during user-testing, refining the intervention and addressing economic considerations. These actions were completed

in preparation for the next stage of the project which would be to move on to the "Feasibility" stage of the MRC Framework.

5.2 Relevant literature

Methodological frameworks exist for designing apps for children, however they are primarily tailored towards education [221]. In 2022, Chen described work which was carried out to develop a framework, with a focus on the emotional needs of children when developing apps [221]. Chen goes on to explain that this is important in the field of educational and entertainment apps for children. Elements of Chen's work are applicable, but a direct focus on healthcare app development is absent. Recent work in the field of healthcare apps for children demonstrates that there are many variables to be considered [222]. Lee et al reviewed child-health apps and produced insights into app development for children such as the consideration of a child's developmental stage, and creating age-appropriate material. Despite this, a robust methodology for the development of self-management healthcare apps for children does not exist.

The lack of robust, child-specific methodology to design, develop and implement digital interventions meant that literature relevant to adult populations was considered. This literature informed the production of the NON-STOP app. The theoretical approaches used to develop the app map to those used across the doctoral programme by the researcher, specifically SDT [78] and SEM [109] which are discussed below. Literature was considered relating to the development and evaluation of complex interventions following the MRC Framework [44], as well as guidance on digital technologies from NICE [223]. The Behaviour Change Wheel (BCW) [145], a taxonomy of psychological theories relevant to supporting behaviour change, was used to identify additional behavioural theories that were to be considered during the production of the NON-STOP app. All are discussed in more detail below.

5.2.1 Theoretical approach

The way in which the NON-STOP app was designed and developed lends itself well to the overarching approach of pragmatism [38, 42], described earlier in the thesis. While aligning with a pragmatic paradigm, the NON-STOP app was informed by intervention development framework and theories in order to address the aims and objectives, this is discussed in more detail in section 5.2.2.

Designing the NON-STOP app required input from multiple stakeholders. These stakeholders included children with Perthes' Disease, family members, clinical specialists and specialists in healthcare app design. For projects like this, the views of many are not easily confined to one specific framework or approach. With this in mind, one must adapt as the requirements of others change. The range of frameworks and approaches considered are outlined in relevant sections below.

The umbrella review, discussed in Chapter two demonstrated that there is no robust evidence to inform selection of digital intervention content [73]. The searches used to develop this review have been repeated (most recently September 2024), no new evidence has been produced. The approach taken to create the NON-STOP app was, therefore, realistic and pragmatic, based on the guidance from experts in the field and other stakeholder groups such as patient and public involvement.

The design of the NON-STOP app aligned with the psychological theory underpinning the programme of work which includes SDT and SEM [78, 104] described in detail in Chapter two**.** Both theories consider the motivation and factors affecting the actions of people. These theories were used to guide qualitative interviews with children, their families and clinicians in Chapter three. In reference to the development of the NON-STOP app, these theories informed the sections of the app that were designed to motivate users to use the app, whilst still providing the skills necessary to complete them and increase their autonomy, i.e. self-management.

The psychological theory underpinning the programme of work, as well as the researcher's epistemological and ontological stance of pragmatism, informed the selection of intervention development methodology.

5.2.2 Approaches to intervention development

The NON-STOP app was developed following a blended combination of approaches, presented in the methodology section in Chapter two. Namely the MRC Framework, and the BCW. This section provides an overview of these approaches, and their influence specifically to the development of the intervention. The development of the intervention was also aligned with the GUIDED checklist for reporting intervention development in health research [224]. These points are seen in more detail in subsequent sections, and a copy of the checklist is attached as Appendix N.

5.2.2.1 MRC Framework

The MRC and the National Institute for Health and Care Excellence (NICE) have produced literature relating to complex interventions and digital health technologies, respectively. This has been discussed in Chapter two. For the purpose of this section, the information is given with particular relevance and context to developing the NON-STOP app.

The MRC Framework allowed a flexible, yet robust approach to intervention development and early acceptability testing. It also provided a clear framework for future research to evaluate and implement the NON-STOP app, beyond the scope of this doctoral programme of work. Table 5.1 maps the core elements of the MRC Framework with examples of activities undertaken relating to the development of the NON-STOP app. There are many examples throughout this chapter on how intervention components were selected based on evidence, their anticipated mechanisms of action, and how this combined into a programme theory for the NON-STOP app. The MRC framework also places emphasis on understanding the contextual factors when developing an intervention. This was important here as the Perthes' Disease is strongly clustered into children from socio-economically deprived backgrounds. Ensuring the intervention was designed to be accessible and relevant to families from a range of backgrounds was important. Here, this led to exploring availability of smart phones (including data costs) in families to ensure that an app would be accessible to them.

Table 5.1 – MRC Framework core elements in context to NON-STOP app

Core element	**Relevance to NON-STOP**
1. How does the intervention interact with its context?	The app was designed with children with Perthes' Disease in mind.
2. What is the underpinning programme theory?	Programme theory developed and refined through the programme of work using logic models [225] developed at key milestones within project.
3. How can diverse stakeholder perspectives be included in the research?	Regular PPI and PAG meetings were held, as well as user-testing sessions. Content input was sought from clinicians.
4. What are the key uncertainties?	The content of the NON-STOP was unknown at the outset. Earlier studies, such as the qualitative study [155] and the clinical consensus study [219], informed the content uncertainties. In addition, the final study, using a mixed-methods approach, preliminarily tested the intervention. It tested uncertainties around acceptability and usability of the intervention.
5. How can the intervention be refined?	An iterative approach to intervention development (i.e. the NON-STOP app) was undertaken, with alpha and beta testing. This was conducted through user-testing sessions, including observations and discussions about app performance. Similarly, following the final study, refinement is possible.
6. Do the effects of the intervention justify its cost?	Economic evaluation was not carried out at this stage. There was consideration of access to smart devices and ability to use at home i.e. self-manage.

5.2.2.2 NICE evidence standards framework

The aim of the NICE evidence standards framework (ESF) [223] is to describe the evidence that demonstrates the value that a digital health technology (DHT), such as the NON-STOP app, has within the UK health care system. The framework allows the developers to assess areas of potential risk and mitigate them through recommendations, often involving demonstrations of effectiveness or behaviour change techniques.

Areas of particular relevance to the development of the NON-STOP app from the framework were reviewed regularly to ensure they met the necessary standards. To give an example, the NON-STOP app fits in to tier 3a Digital Health Technology due to its self-management and behaviour change components. A detailed rationale for the classification has been outlined in Chapter two. Best practice for this tier when looking to demonstrate effectiveness is to consider certain outcomes. These should include patient reported outcomes, evidence of positive behaviour change, and user satisfaction. In the context of the NON-STOP app, it was developed with the potential to measure these by incorporating things such as the progress monitor and rewards.

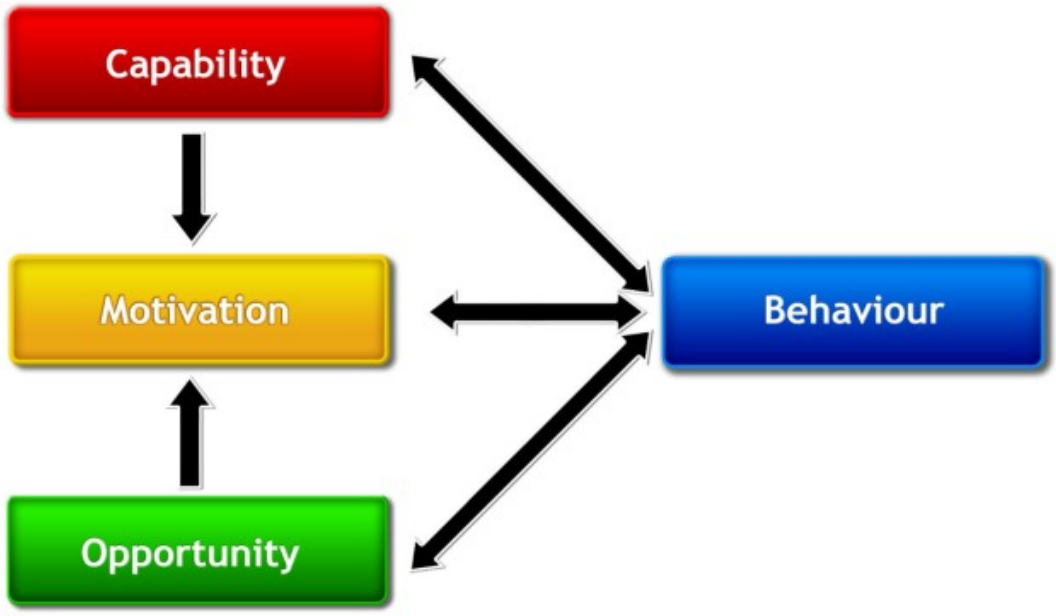
The risks of digital health technologies are cumulative. For instance, the NON-STOP app sits in tier 3a due to its potential ability to help children with Perthes' Disease self-manage. It should, therefore, meet the evidence standards of tier 3a, but also of tiers 1 and 2. Key elements of the earlier tiers include user-involvement in design and testing, as well as experts in the field. The risk in the NON-STOP app was minimised by ensuring that the PAG and PPI engagement as well as user-testing processes were extensive. By ensuring that the app was designed with the viewpoints of those who would use it, ensured that this risk was minimised.

Behaviour Change Wheel

The BCW has been presented in more detail in Chapter two. In the context of the NON-STOP app, the COM-B model (Figure 5.1) aligns well with one element of the theoretical underpinning applied in this doctoral programme of work, the SDT [104]. In SDT, motivation is a key factor considered when theorising what must take place

for an individual to carry out a behaviour. To compliment the work done to develop the NON-STOP app, the researcher underwent post-graduate training in the BCW approach to increase understanding of behaviour change techniques within interventions. Whilst it was not the sole method of intervention development/evaluation, the learning supported the influence BCW had on the development of the NON-STOP app. Using the BCW enabled the researcher to highlight the behavioural target which was to have children regularly complete physiotherapy exercises and engage with self-management elements such as education. From this the researcher could identify intervention functions such as the learning section in the NON-STOP app. Another example is incentivisation as an intervention function. Finally these were integrated as techniques in the NON-STOP app as functions within, such as rewards for completing exercises, reminders to complete and progress tracking.

Figure 5.1 – The COM-B system*



Reproduced from Michie, S., M.M. van Stralen, and R. West, The behaviour change wheel: A new method for characterising and designing behaviour change interventions. Implementation Science, 2011. **6(1): p. 42.*

A primary function of the NON-STOP app is to support behaviour change, i.e. to use the app for self-management by making it usable for the children with Perthes' Disease and their families. The app is specifically designed to optimise their

capability, and in turn increased the likelihood of the behaviour taking place. Similarly, the **motivation** to use the app was addressed with things like incentives for completing their exercises and making the app fun and interesting to the users. Regarding the **opportunity**, the app was made available to participants for them to use on their personal smart-devices, and designed to encourage self-management behaviours wherever they saw fit.

The BCW intervention functions (the middle layer, Figure 5.2) are then linkable to behaviour change techniques (BCTs). These BCTs were organised into a taxonomy, grouped by experts in a Delphi-type exercise in 2013 [226]. The author group was once again led by Michie, and developed a taxonomy of 16 groups of 93 BCTs. Whilst the BCW and BCT mapping approach was not fully implemented for the NON-STOP app, elements of the NON-STOP app were mapped to specific BCTs. Table 5.2 provides some illustrative examples of how specific BCT categories were used, and a practical demonstration of how this was done in the NON-STOP app. To give an example, in BCT category ten, "Reward and threat". The techniques within this category relate to rewards and incentives to increase the likelihood of the behaviour taking place. In the NON-STOP app, an example of a reward suggested by participants in the qualitative study was of an 'in-app' rewards, in the form of stars given to the user every time they hit their exercise goal. Stars could then be used to 'purchase' customisation options for their personal avatar. In turn, motivating them to repeat the behaviour. A potential limitation in not applying a full systematic BCW mapping is that there may not be full coverage of all behavioural determinants that might impact the uptake/use of the NON-STOP app. This process is time-consuming, and beyond the resources available in this thesis. The selection of relevant BCTs was done based on evidence, theory and stakeholder input, which was necessary to expediate the lengthy BCW mapping process.

Table 5.2 – Behaviour change techniques applicable to the NON-STOP app

BCT Category	**Specific technique within category**	**Examples of how this is addressed in NON-STOP app**
1. Goals and planning	1.1 Goal setting (behaviour)	Users given goal of how many times to use app each week

	1.2 Problem solving	If a user selects a pain score of "8-10 Hurts a lot" then they are suggested to review the wellbeing information in the Learning section
	1.6 Discrepancy between current behaviour and goal	Users encouraged to complete set time per exercise and attention drawn to this if not met e.g. aim of 30 seconds of stretching but only achieving 10 seconds.
2. Feedback and monitoring	2.3 Self-monitoring of behaviour	Activity diary for users to log when they have completed exercises
3. Social support	3.1 Social support	Advice on app about accessing online groups open to children with Perthes' Disease and their families (STEPS charity)
4. Shaping knowledge	4.1 Instruction on how to perform the behaviour	On first load of the app, users receive instructions on how to use the app. Instructional videos of how to perform each exercise on the app.
5. Natural consequences	5.1 Information about health consequences	In the information on the app, users are told the consequences (getting stronger) of doing exercises.
7. Associations	7.1 Prompts/cues	Push notifications for users to log on to the app and use to reach their weekly goal
8. Repetition and substitution	8.7 Graded tasks	Initial goal is to use the app three times a week, working up to five times a week over the six-week testing period.
10. Reward and threat	10.8 Incentive	Instructional demonstration of the app informs users of their goals and the reward (stars to customise the avatar).
	10.10 Reward	Users get stars for completing their exercise goal per day as they use the app.
12. Antecedents	12.4 Distraction	Strengthening exercises are designed in a fun way e.g. jump squat being delivered as a "frog jump" with cartoons

	12.6 Body changes	Prompts for strengthening and stretching exercises, as well as education, about the benefits of doing these (stronger and potential for reduced pain)
14. Schedule consequences	14.9 Reduce reward frequency	Stars are gained for completing exercises three times a week initially but increases to five times a week by end of app-testing period
15. Self-belief	15.1 Verbal persuasion about capability	Learning section, as well as instructional demonstration, explains that elements of the app like exercises and wellbeing can be done despite pain/diagnosis

Components of the NON-STOP app and relationship to BCW

The NON-STOP app can be divided into five key elements that contribute to the self-management of Perthes' Disease, including:

1. The **Learning section**, where information is stored about Perthes' Disease, how the app works, and other components such as wellbeing guidance;
2. The **Avatar**, that users personalised as they continued to use the app;
3. A **Progress section, including an activity diary** for users to log their daily/weekly use of the app and monitor their progress as well as their pain levels.
4. The **Activities section** contained the strengthening and stretching exercises for users to complete.
5. **Push notifications** delivered to users to provide a prompt for them to use the NON-STOP app and achieve their weekly goal to earn rewards.

In Table 5.3 the key elements of the app are described with some examples of how they relate to the three theoretical models/approaches (BCT, SDT and SEM) underpinning of the NON-STOP app. One example from SDT relates to how changes in behaviour over time may lead to sustained changes in behaviour such as habit formation. An influencing factor can be intrinsic motivation, meaning that an individual may gain satisfaction from the desired behaviour rather than the

behavioural techniques that drive the behaviour. To give an example for the NON-STOP app, rewards are given to customise the avatar, which may motivate children to engage initially, however if over time, they do more exercises, and have less pain due to this, the motivating factor becomes engagement with the intervention as opposed to exercising purely for rewards.

Table 5.3 – Key elements of the app and how they map to each of the theory elements

App component	BCT	SDT	SEM
Learning section	Shaping knowledge	Competence and autonomy	Self-efficacy that comes with self-management
Avatar	Reward and threat (incentive)	Autonomy and relatedness	N/A
Progress section	Feedback and monitoring (self-monitoring of behaviour)	Autonomy	Self-efficacy that comes with self-management
Activities section	Antecedents (distraction and body changes)	Competence, autonomy and relatedness	Child (age, self-efficacy) and interpersonal (family and peer involvement)
Push notifications	Associations (cue signalling reward)	N/A	Interpersonal (family support with reminders)

5.3 Programme theory and use of staged logic models

Programme theory describes how and why an intervention is expected to work, detailing the processes, mechanisms of action and outcomes that link the intervention activities to its intended effects [227]. Logic models are a visual representation of the programme theory. Tools used to display and describe the mechanisms of action within an intervention illustrating its programme theory [228]. Using logic models, it is possible to demonstrate the factors that influence the

intervention with a focus on inputs, outputs, change mechanisms, measuring change and impact. Logic models also take in to account contextual factors such as resources and contributing strategies/policies. It is possible to use logic models as a tool for understanding what factors influence any potential change. In the context of the NON-STOP app, these factors provide a visual representations of how each factor influences change for the children with Perthes' Disease and their families.

Frequently in practice logic models are fixed. They are produced at the first stage of intervention development and used to evaluate the effectiveness of the intervention. It has however been suggested that more flexible and dynamic models are required whereby the logic model is updated as new evidence is accrued [229]. An iterative approach to logic models was used in this project, a summary is provided below, and in Table 5.4.

A preliminary logic model was produced with input from a methodological expert in the supervisory team (SR). The preliminary logic model was created after the evidence synthesis stage. It was then reviewed at two key milestones: after the completion of the qualitative study in Chapter three, and following the clinical consensus study in Chapter four, and the feasibility study in Chapter six. The later iterations of the logic model were reviewed by the wider supervisory team, integrating clinical and theoretical understanding of the logic model. After the final study of this programme of work, the logic model was once again reviewed, and the post-intervention testing logic model was produced. The final logic model incorporated input from the clinical consensus study and further PPI conducted prior to the final study. The rationale for the iterative approach was to ensure that the project was supported by programme theory, as recommended in the MRC Framework. Emphasising the importance of continually reviewing contextual factors.

All three logic models can be seen in Appendix I, however in Figure 5.2, the final logic model demonstrates the programme theory which incorporates sources of information gathered throughout this project and maps to how the information (or inputs) could lead to improved self-management for children with Perthes' Disease. The mechanisms of action theorise how the intervention components can lead to the desired outcomes. For example, the clinical consensus study highlighted the need for

hip strengthening that can reduce pain for the patient. The consensus study also highlighted the need for education relating to Perthes' Disease which aligns with the psychological theory (SDT) that suggests through increased competence, an individual will be more motivated to change a behaviour, and sustain the change.

Table 5.4 – Logic model summary

Logic model title	**Time-point**	**Content**
Preliminary logic model	Pre-qualitative study	Evidence available prior to qualitative study [28, 34] and input from PPI. Plans for future work and potential impact.
Interim logic model	Post-qualitative study	As above, with additional information from qualitative study [155] and plans for intervention development.
Final logic model	Post-intervention testing study	As above, with additional information from clinical consensus study [219], further PPI and app-testing trial.

Figure 5.2 – Final logic model after post-intervention testing study

Final logic model NON-STOP Logic model v3

Problem addressed

Based on current evidence, there is a need for the development of a digital self-management intervention for children with Perthes' Disease.

Priorities for intervention

1. *Previous case review highlighting significant variation of 'usual' non-surgical management for Perthes' Disease in the UK [1]*
2. *Based on systematic review that found no robust evidence to support the use of any non-surgical intervention compared with another [2]*
3. *Digital intervention ('app') designed using evidence-based guidance (MRC, PPI, theoretical input) [3] qualitative interview study [5] and content from clinical consensus study [6]*
4. *James Lind Alliance identified need for research exploring outcomes following non-surgical care of Perthes' Disease [4]*

Inputs	Outputs (activity)	Change mechanisms	Measuring change	Impact
Children/families - Participants following self-management support - Videos/examples of exercises to be used - Involved in the JLA priority setting for this topic - Access to smart phones - Qualitative work highlighted importance of inclusion in decision making Clinicians - Children/families regularly reviewed in outpatients - Input to identify important content for app - Training staff on app use - Clinicians' training children/families to use app - Empowering patients important to clinicians (from qualitative work) Intervention (the app) - App developers' expertise on 'what works' in app design - Empowerment and patient control important (from qualitative work)	App design - Early versions of app, including training for app, led by developers. - Clinical content of app informed by an umbrella review & clinical consensus work (Delphi study) - App design tested iteratively - Clinical content from consensus study Early testing of app - Clinicians trained on how to use/support children/families - Children/families test out app (acceptability, feasibility of use). Metrics of use gathered. - Look to utilise PPI groups (PAG & YPAG) to do some informal testing of app – usability and acceptability	- Consensus study identified change mechanisms supporting to maximise engagement with app - Self-determination theory: Changes in 'psychological needs' – increasing the motivation of the child/family to complete their physiotherapy - Exercises will strengthen and lengthen relevant muscles and in turn, stabilise hip joint - Educational/'learning' elements from the NON-STOP app also provide motivation through increase in competency and autonomy of individuals	Process measures - Metrics from app measuring usage (how many times used, how long for, what has been accessed when logged on). - Qualitative feedback on use of app from children/families and clinicians Outcome measures - PROMs from Core Outcome Set (PROMIS mobility) - Radiological outcomes (Stulberg or equivalent) - Clinical markers such as ROM and strength. Pain captured in PROM - Usability outcomes specific for digital health technologies.	- App that can promote improve self-management behaviour - Early data relating to engagement with a novel intervention for self-management - Improved clinical and PROM outcomes - Potential to reduce need for surgical intervention - Implementation within clinical trial as part of non-surgical intervention

Contextual factors

Resources
- Remote self-management increasingly common in rehab settings post-COVID.
- Time for children/families to do the exercises/aspects of the app.
- Time for clinicians to train children/families on how to use the app.
- Access to internet/smartphone to use the app
- Could app be used outside of family settings e.g., in schools, or other activities?

Service
- Some centres don't have specialist clinics where these patients are 'located' clinically. So could lose out due to difficult making clinicians aware of intervention. Although qualitative work did not highlight any obvious issues

1. Galloway, A.M., et al., *A case review to describe variation in care following diagnosis of Perthes' disease.* BJO. 2020. **1**(11): p. 691-695
2. Galloway, A.M., et al., *A systematic review of the non-surgical treatment of Perthes' disease.* BJO. 2020. **1**(12): p. 720-730.
3. Skivington K, Matthews L, Simpson S A, Craig P, Baird J, Blazeby J M et al. A new framework for developing and evaluating complex interventions: update of Medical Research Council guidance. BMJ. 2021; 374: n2061 doi:10.1136/bmj. n2061
4. Vella-Baldacchino, M., et al., *Research priorities in children requiring elective surgery for conditions affecting the lower limbs: a James Lind Alliance Priority Setting Partnership.* BMJ Open, 2019. 9(12): p. e033233
5. Galloway, A.M., et al., *"Waiting for the best day of your life". A qualitative interview study of patients' and clinicians' experiences of Perthes' disease.* Bone & Joint Open, 2023. **4**(10): p. 735-741.
6. Galloway, A.M., et al., *Clinical consensus recommendations for the non-surgical treatment of children with Perthes' disease in the UK.* The Bone & Joint Journal, 2024. **106-B**(5): p. 501-507.

5.4 Developing the NON-STOP app

To understand the stages of app development, this section outlines the early stages including procurement and contracting of an external app development company. After this, an overview of content development and user-testing processes are described.

5.4.1 Procurement and contracting process

As part of the researcher's doctoral programme of work, funding was secured to have the physical aspect of the NON-STOP app created by an external provider. This relates to the creation of the programming and launching on app-stores, for instance. Prior funding allowed the researcher to conduct exploratory work. PPI activities as well as input from others who had experience of digital interventions identified the need for expert services for app development.

In the early stages of this PhD, work took place to explore potential providers, and gain an understanding of what providers could bring to the project for app-development. This exploratory work involved working with NHS Digital, to discuss the stages of app design, development and testing prior to any research studies or implementation. NHS Digital offered valuable insight in to their experiences of creating digital interventions, and offered their experience in the field of child-health digital interventions. It is here that the researcher was put in contact with HMA Digital Solutions (HMA).

As part of the development of this project application, there was a consideration of whether sub-contracting was required or whether in-house designers/development could be utilised within University of Leeds. However, HMA have previous experience of working with research projects completed by doctoral and post-doctoral fellows. As a company they understood the difference between creating a product independently for use, and designing and developing an intervention iteratively and collaboratively in preparation for testing in a research study. Their experience also spanned across the child-health sector, including a digital self-management

intervention for children and young people with Diabetes [230, 231]. HMA also have experience of working with the University of Leeds, and the National Institute for Health and Care Research (NIHR), funders of the researcher's fellowship.

There is also available literature that outlines optimal providers for things such as app-development in clinical research. In 2022, Chettri et al explained that researchers should seek those with experience related to research and implementation rather than those solely focused on proof of concepts or demo-versions of apps [232]. The authors summarised their challenges and explained that in their future work they were planning to build a team that had expertise in domains of app-design. They considered user-interface, security and ensuring that apps are native to their operating system (Apple's iOS or Google's Android). It is important to understand it was not realistic within the scope of this project for the researcher to gain the necessary expertise in app design and development. Nor was it the aim of this doctoral programme.

In order to meet the requirements of the University the tendering process was reviewed, and advice from the procurement team was sought [233]. After some discussions, and whilst complying with the University of Leeds guidance, it was agreed HMA were the optimal providers. Approval was gained and the contracting process was completed.

5.4.2 Influence of previous work

As well as the input from key stakeholders and PPI/PAG, the NON-STOP app was informed by the studies that took place in the earlier part of the doctoral programme. A brief overview of each is given below, with a particular focus on how each empirical study contributed to choices relating to the app content and delivery.

Table 5.5 below outlines the sections of the NON-STOP app and the elements of the project that contributed to the content.

Table 5.5 – Sections of the app and the contributing elements of project

App section	**Contributing elements of project**
Activities	Qualitative study – strengthening exercises were fun and based on characters. Example: frog jumps instead of jump-squats. Users also are given a choice to complete exercises they want to, relates to the theme of children with Perthes' Disease and their families being key decision-makers in their own care. Clinical consensus study – exercises were focused on hip ROM and strength.
Learning	Qualitative study – All stakeholders explained that understanding was important to them. Clinical consensus study – elements of the learning section such as wellbeing and education around the disease process achieved consensus in Chapter four.
Progress	Qualitative study – the idea of an avatar/character that was customisable came directly from the qualitative study from children with Perthes' Disease, with character designs coming in user testing and PPI sessions. They spoke of levels and rewards being motivating to them.

Qualitative study

In the qualitative study [155], interviews with children with Perthes' Disease, their families, and clinicians explored their experience of previous care and opinions towards a digital self-management intervention. The main findings relating to the digital intervention were that stakeholders wanted accessible information allowing them to understand their condition and how to manage it. In the qualitative study, as well as in PPI work completed, participants identified that a smart-device application was an acceptable intervention delivery mechanism. All qualitative study participants agreed that there was a strong need for consensus on management approaches to inform non-surgical treatment of Perthes' Disease. Children suggested specific content they would like to see in the NON-STOP app, including customisable avatars and rewards that the children 'earn' by using the app. This was a key step in the app-development. The qualitative study findings therefore supported inclusion of an education/information section ('Learning section') in the app.

Clinical consensus study

The consensus study was a modified Delphi study, described in Chapter four, to achieve clinical consensus on the non-surgical treatment of Perthes' Disease [219]. The clinical consensus study informed the clinical content of the NON-STOP app. The intervention functions were identified by gathering expert opinion on what non-surgical treatment of Perthes' Disease needed to include. The aim was to develop an intervention that would lead to a change in behaviour i.e. children completing hip exercises and engaging with educational content. For example, in the Activities section of the app, the exercises chosen for inclusion included those around hip stretches and strengthening. Both of these exercises achieved clinical consensus in the Delphi study to say that children with Perthes' Disease, irrespective of stage or severity, should complete stretches and strengthening exercises for the hip.

After the multitude of work that went into its creation, the NON-STOP app consisted of four main components. The components of the app were influenced by behaviour change techniques to try and achieve these, such as goal setting and remainders. These are described below with a focus on their creation and considerations for how the section of the app was designed and developed to meet the needs of the users.

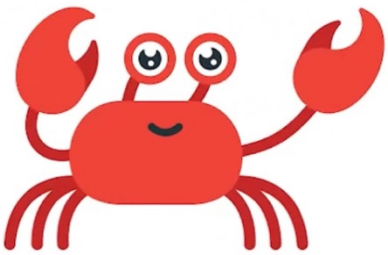
5.4.3 Activities section

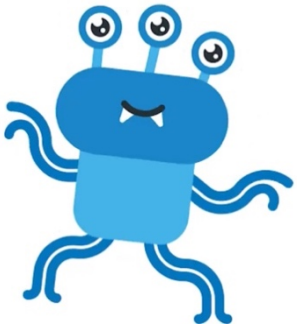
Typically children's physiotherapy includes making exercises, activities and stretches relatable and fun for the patient. This is commonly done by integrating play into therapy sessions, often by using characters or animals. For instance, in the NON-STOP app, calling squat jumps, that exercise all muscle groups in the lower body, "frog jumps". Incorporating play and gamifying physiotherapy is commonly used. The benefits of this 'gamification' of physiotherapy has been well documented in conditions that rely on physical activity for long-term management, such as cerebral palsy [234] and cystic fibrosis [235]. The clinical consensus recommendations stated that lower-limb strengthening should focus on hip strengthening and that ROM exercises should focus on maintaining abduction, rotation and flexion/extension of the hip. Activities were selected for the app accordingly. If users logged in and used

the NON-STOP app at least once a week for the first four weeks, they unlocked two additional exercises. These exercises were frog jumps and scissor jumping. Due to the jumping involved, these exercises (jump squats and jump lunges) are considered more difficult, and so were deemed appropriate to introduce later in the testing period, once user-ability had improved. This element of 'unlocking' activities was included in the NON-STOP app to explore whether app use was frequent enough to result in the exercises being unlocked rather than any clinical change.

Once the activities were selected, making them 'child-friendly' was the next step. All were to be delivered as videos. The strengthening exercises were simple enough to alter to incorporate into cartoon characters for the app based on commonly used descriptions of the actions. A more detailed description of all of the strengthening exercises included in the NON-STOP app are presented below in Table 5.6.

Table 5.6 – Exercises in the 'Activities' section and their origin

Exercise	**Action**	**Name for app (and character)**	**Image used**
Crook-leg fall out	In crook-lying (knees and hips at 90 degrees), allow the knees to fall out whilst keeping ankles together, then bring together slowly.	Butterfly wings (butterfly)	
Squat walking (sideways)	In a squat position, walk sideways the length of a room, and whilst maintaining the squat position, walk sideways back to your starting point.	Crab walk (crab)	

Squat walking (forwards/ backwards)	In a squat position, walk forward the length of a room, and whilst maintaining the squat position, walk backwards to your starting point.	Monster walking (monster)	
Sideways squats	In a sideways squat, squat down, return to neutral, then squat to the opposite side.	Ninja squat (mouse)	
Jump lunges	From the lunge position, jump and move your legs so the foot that was at the front is now at the back, and vice versa.	Scissor jumps (cat)	

Jump squats	Squat, on the return to neutral, jump and land into another squat.	Frog jumps (frog)	

As well as creating the cartoon character videos for the strengthening exercises, it was important to make sure that there was a clear, accurate instructional video for participants to use to learn how to complete the activity. To do this, a motion capture effects were used to turn human demonstration into a cartoon that demonstrated accurately how to complete the activity. This was accompanied by text describing how to complete the activity and can be seen in Figure 5.2 below.

Figure 5.3 – Examples of the descriptive instructions for activities

It was possible to transform exercises to a cartoon character due to the nature of the activity. There were elements that required careful consideration, such as the stretching exercises. It was not possible for the stretching exercises to be cartoon characters, like the strengthening exercises, because most had a passive element that required a family member to perform. In order to demonstrate this clearly in the app, a green silhouette of a human was used to demonstrate the position to hold the child with Perthes' Disease. Examples of this are shown below in Figure 5.3.

Instructional videos and timers for completion were created by the designers from HMA. Both in the cartoon character version, and an avatar of a human that ensured accuracy as well as the fun element of the activity. The avatar and silhouette were

created using digitalisation of a human carrying out the activities (often the researcher and a member of the HMA team).

Figure 5.4 – Examples of pages in the Activities section

There are other aspects of the Activities that were built in to the coding of the NON-STOP app. Elements that are ultimately safety features, whilst not a true risk of 'harm' to the child, built in to provide a level of assurance for the researcher and the team that use is tailored to the needs of the child. To demonstrate this, consider the stretching and strengthening exercises included. The clinical consensus recommendations suggested that both hip stretches and strengthening should be part of the non-surgical treatment of Perthes' Disease. With this in mind, the app developers built in a code so that users could not start their programme until they had selected at least two stretching and two strengthening exercises. They could select more, but not less as seen in Figure 5.3 the rationale for this feature was to maximise a 'thoroughness' of children completing physical activities that both stretched and strengthened muscles.

Similarly there were considerations also built in regarding he pain levels of app users. The Wong-Baker scale was built in to the NON-STOP app, and is a validated outcome measure for measuring paediatric pain [236, 237]. The measure is an ordinal

assessment of pain outcomes, using a series of six facial-expressions to illustrate the degree of pain intensity [238]. A numerical rating is assigned to each face (from 0 - 'no hurt' to 10 – 'hurts worst'). It has been validated for use amongst children over 3-years-old [236]. This tool had a particular strength in this app-testing study because it could be used by all participants in the study as it is validated for self-reported use from 5 years old [239, 240]. It is particularly useful in children, as only one third of children up to 14 years understand the concept of a visual analogue scale [241]. The Wong-Baker FACES Pain Rating Scale has been demonstrated to be useful in older children aged 8-17 years old, correlating closely with other pain tools, such as the Visual Analogue Scale [236]. This measurement for pain has not been used in studies involving children with Perthes' Disease, however it has been used in previous children's orthopaedic studies. In these studies it produced almost identical scores amongst children <8 years and those aged 8-16 [242]. This was integrated in to the NON-STOP app as shown in Figure 5.4.

Figure 5.5 – Adaptation of the Wong-Baker scale for NON-STOP app*

** Wong-Baker scale adapted from, Garra., et al., Validation of the Wong-Baker FACES Pain Rating Scale in pediatric emergency department patients. Acad Emerg Med, 2010. **17**(1): p. 50-4.*

Another example of where the NON-STOP app had built-in safety features. If a child completed their daily pain score and had scored 0 or 2 on the Wong-Baker faces pain scale, they were advised to complete their exercises and aim for 60 seconds. If they had completed their pain scale score for the day and scored 8 or 10, they were only advised to aim for 30 seconds.

5.4.4 Learning section

The learning section aimed to address the needs of the users that were discussed in the qualitative study [155]. In the interview study, key stakeholders expressed their desire for accurate information, readily available to them. The learning section was built to comprise key elements of education that were felt to be necessary for children with Perthes' Disease and their families, based on the results of the clinical consensus recommendations study [219]. The Learning section was also informed by psychological theory, as it was intended to increase the autonomy of the user which is a key part of the SDT.

The NON-STOP app needed to be clinically robust in terms of accurate and reliable clinical content. In the absence of robust evidence for non-surgical treatment of Perthes' Disease, the clinical consensus study provided the best alternative. Consequently, the Learning section of the app includes clinically accurate advice and information on the anatomy, physiology, and typical disease process of Perthes' Disease. For instance, the consensus study highlighted the importance of providing children with Perthes' Disease and their families with information about the disease process, including anatomical changes. This is included in a page called "What is Perthes' Disease?" in the Learning section of the NON-STOP app. Information about the condition, amongst other aspects of care, can be seen in Figure 5.5.

All content of the Learning section was written to be understandable by children and adults, and was written to a reading age of five years old. It was assessed for appropriateness using the Flesch-Kincaid Grade Level system [243] and reviewed by PPI groups. Table 5.7 summarises each of the eight sections' content.

Table 5.7 – Summarising the content of the Learning section of the NON-STOP app

Learning section	**Content**
What is Perthes'?	Information to give an understanding of what Perthes' Disease is, how long the disease process lasts and what the aims of treatment are. External link to a charity website (STEPS) that the researcher and members of the supervisory team (DP) contribute [244] to.
Nutrition	Information about the importance of a balanced diet. Link to UK national guidance for nutrition for children [245].
Lifestyle	Advice on the levels of daily exercise and activity that children with Perthes' Disease should achieve. External links and graphics demonstrating this [246]. There is also information relating to activity modification which is common in Perthes' Disease.
Gait	This section outlines the rationale for why and when children with Perthes' Disease may need to alter their mobility using things like walking aids or reducing the amount they walk.
Research	In the research section there are YouTube links for recent webinars relating to research currently taking place in Perthes' Disease. There is also a link to the NON-STOP project web-page [247].
Pacing	Activity diaries are discussed in the pacing section with information for users about how to plan their weekly activities based on things like pain levels and recovery time.
Pain relief	The pain relief section covers the information gathered in the clinical consensus recommendations study relating to pain relief. This includes advice relating to medication (as per packet instructions) and using other methods of pain relief such as hot/cold therapy.
Wellbeing	The wellbeing section gives users information about the impact of Perthes' Disease from a psychosocial perspective. There is information in this section about using the STEPS charity website [244] and the NHS website for advice on managing a child's wellbeing [248].

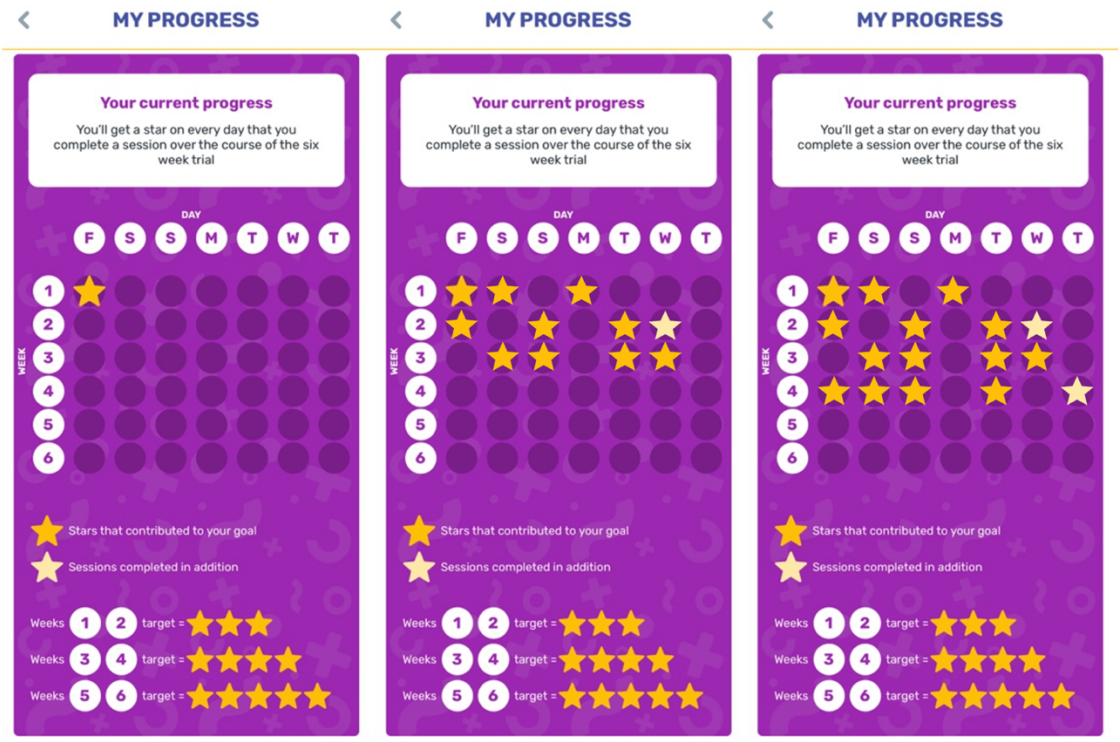
Specific to the design of these sections, elements of the topics relied on links out of the app. For example, in the research section, there was originally a link to a YouTube video of a webinar delivered by the researcher and one of the supervisory team. This webinar was provided to STEPS charity to provide an update on research currently being completed in the Perthes' Disease population. Initially this was displayed as text with html underlined in blue writing. This was considered to be sub-optimal in terms of appearance on the app. Instead, a change was made so that a thumbnail image of the YouTube video was used with an embedded link that took the participant to the YouTube video. A caption with the instruction to click the thumbnail was used. This was repeated in other sections of the Learning section where necessary to optimise the visual aspect of the app.

Figure 5.6 – Examples of pages in the Learning section

5.4.5 Progress section

In the Progress section users were able to see how many days they had completed an exercise session that week. In an attempt to mimic progression of exercises in terms of frequency, users had to complete more sessions as the weeks went on to gain the stars. It was important to ensure users were able to do more if they wanted, and whilst it wouldn't contribute to their 'reward' (customising the avatar), it would still be visually displayed. A star would appear but with a slightly different colour, as seen in Figure 5.6, to demonstrate they had completed a session.

Figure 5.7 – Examples of pages in the Progress section



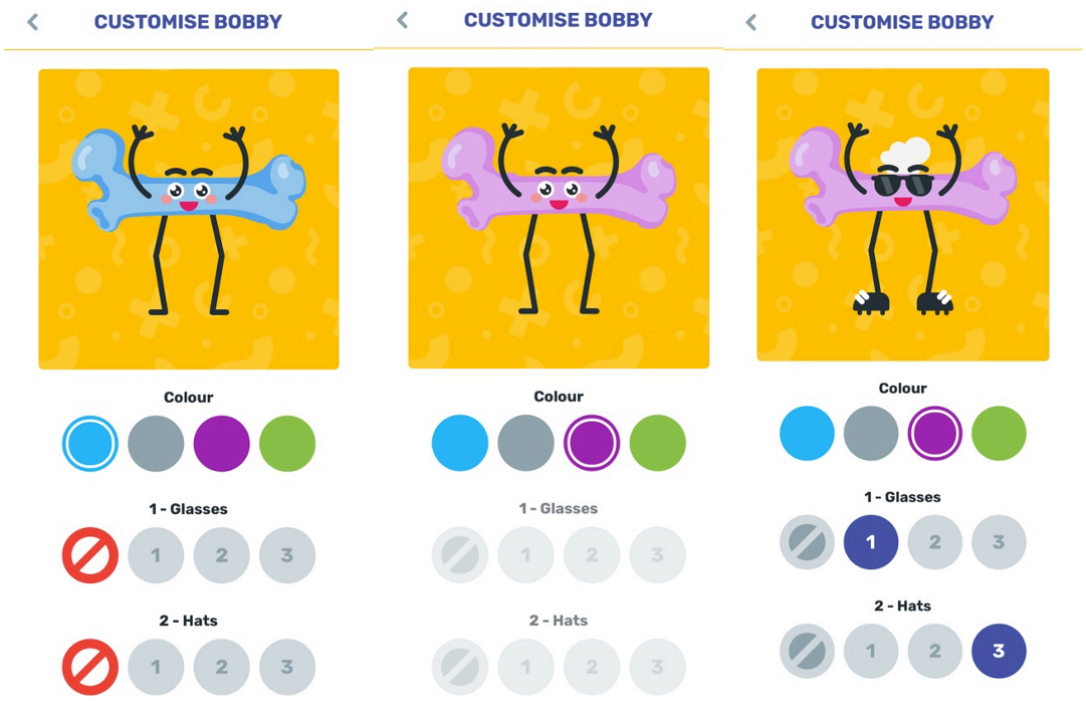
5.4.6 Avatar

The avatar, named "Bobby the bone" by a member of the PAG in the application stages of this programme of work, was integrated fully into the NON-STOP app. The name of the avatar was suggested by a member of the project advisory group however did go through iterations. Initially the avatar was called "Bob the bone man". After review with PPI and PAG, a gender-neutral name (Bobby) was agreed upon, so as to represent all potential app-users.

As outlined, PPI/PAG input over the course of the app development process was instrumental in the avatar section. Being able to customise the character to look how the user wants Bobby to look was key. It was something that was suggested in PPI/PAG sessions, but a customisable avatar that was familiar to the users was also a common theme in the qualitative study from Chapter three. With this in mind, one app development session took place with HMA and members of the PAG where ideas for customisation options for Bobby were discussed. It was in this session that it was suggested that as the six-week testing period progressed, Bobby could earn outfits in 'sections' of their appearance as rewards for engaging with the intervention. Different options for glasses, hats, activities and others were made available and designed by the app development company based on the input from PAG members and can be seen in Figure 5.7.

The design of the NON-STOP app in terms of being colourful, interactive and having a simple layout and structure were based on the findings from the qualitative study as well as PPI/PAG input. It was important to promote a fun and engaging interface in an attempt to promote use. It also aligns with the theories which underpin the project, namely SDT which relies on autonomy and relatedness as a factor influencing motivation. Having Bobby the bone customisable to the user has the potential to make individuals feel represented. Similarly, fun whilst still educational elements in the Learning section ensure accurate information is delivered in a way that is accessible and relatable to the use.

Figure 5.8 – Examples of pages in the Customise Bobby section



5.5 User testing stages

Over the course of the NON-STOP app development, it was necessary to understand the stages of development that are used in the development of digital interventions. These are typically broken down into two stages, alpha testing and beta testing [249]. In an overview given by Naeem et al in 2015, lessons learned from the pharmaceutical industry outlined that prior to the alpha testing stage is a 'pre-alpha' stage in which the software and content are designed. In the context of the NON-STOP app, that is what has been described above. Once the app was designed and developed, alpha testing took place.

Alpha testing of the NON-STOP app happened in a very pragmatic, logical sense. These included members of the app-development team, the researcher, volunteers such as clinicians and members of the PAG. All of whom tested the practical features of the app. This process is in line with what has been done in other alpha testing phases of a healthcare app [250]. Decisions throughout the intervention

development phase were discussed during the sessions by app-users, developers and the researcher. Modifications were discussed and agreed upon within that group. The changes during development were largely practical and presentational. For example, the phrasing that came on the screen in a pop-up box before asking for a pain score had adaptations to the layout and how much text was displayed.

Alpha testing activities are led by the app developers and are a routine part of digital technology development. These activities, as described, are synonymous with stakeholder engagement activities within the digital health technology space. No user data is collected, and for that reason ethical approval for these stages is not required.

In this stage for the NON-STOP app, each domain of the app was used, and specific sections of the app were explored. Specific actions such as selecting a high/low pain score each day to ensure the triggers for wellbeing advice/pacing were given for higher scores. Similarly there are requirements that must be met to gain rewards such as using the app three times a week in the first and second week. This practical use of the app was part of the alpha testing phase to assess elements of functionality. At this point, accessibility issues such as digital inclusivity were explored. These are discussed in more detail in the discussion section of the chapter. Mainly the considerations were around the need for internet access to use the NON-STOP app.

The beta testing stage of any digital healthcare app requires end-users to engage with the app in a 'real-world' setting. This allows the developers and the wider team to identify issues and rectify/adapt the software prior to the release in any full scale [251]. For the purpose of the NON-STOP app, the beta testing is the app-testing trial that follows in Chapter 6**.** At this stage, the testing does follow a protocol and was led by the researcher. For this part of the project, ethical approval was sought, and is described in the next chapter.

5.6 Patient and Public Involvement

Patient and public involvement was vital to the success of this project both in the application stages and throughout the duration of this doctoral programme of work. There were regular sessions in the lead up to application which is where the decision to have the intervention be an app rather than a website/other pathway type of intervention. This idea was suggested by a NIHR Young Person's Advisory Group (YPAG), and thought to be a more amenable method of delivering a digital intervention.

There were regular meetings as per the plan for the NON-STOP project, with visits to YPAG at numerous centres in the UK to discuss the development of the app. However much of the alpha testing and design took place as part of the NON-STOP PAG in collaboration with the app-developers. During these activities, the developers lead the design of user testing sessions given their expertise, the researcher's role was to engage with the users during the sessions.

Sessions spanned from the design process with drawings and ideas for avatar customisation options through to physical user-testing sessions. In the user-testing sessions, PAG members logged on to the NON-STOP app and went through the sections of the app to test how easy it was to load the app, navigate through the sections and discover any areas for amendment. A key example of the effect that these sessions had relates to the avatar customisation. During an app design session, there were suggestions of things that may motivate children with Perthes' Disease to hit their exercise goal. These were things like customising the avatar to look like sports characters that are relevant to children. This perfectly demonstrates the need for users to be involved in the design and development of an intervention that is made for them.

5.7 Training package for users

Users required a training package that starts from the outset when they consider using the intervention. The training needed to include detailed instructions on how to download and register the app included as part of the study sign up. Users also required instructions on how to engage with the app successfully. A training package, suited to their needs, was created and packaged within NON-STOP as “Your journey”.

Another fundamental design choice made during the development process related to the content and delivery of a training package for using the NON-STOP app and specifically if the app was designed to be ‘standalone’ as opposed to requiring an induction by an instructor (e.g. a researcher or clinician). The discussions focused on around whether/how instructional sessions with the researcher and participants (either face to face or over video call) were needed. An instructional session would have involved the participant being shown how to use the app and talked through the sections of the app. Through PAG discussions, as well as discussion with the supervisory team and app developers, a decision was made to create the “Your journey” section of the app to provide the same information as any 1:1 training session for participants. Providing the training package during the app onboarding was also a positive when considering the potential future rollout of the intervention. It is much more sustainable and efficient to deliver training packages for users this way rather than face to face training with a clinician.

When users logged on to the NON-STOP app for the first time, the app was designed so they could not progress to using the app without first viewing the “Your journey” information section. The section walked the participants through the app, demonstrating the various sections of the app as well as outlining the aims over the course of the testing period. The final part of the training package information informed participants that the “Your journey” could be viewed at any time in the future by selecting it from the menu in the corner of the screen. This allowed participants to review instructions should they need to at any point in the testing period. Minimising burden to both the participants, and the researcher should any issues arise.

5.8 Safety/support

It was important to consider confidentiality and protect highly sensitive personal data within the app use. To maximise this, information was limited to a single email address per participant given to HMA to allow users to register and download the app. After this, the users registered using a user ID chosen by them as well as a 4-digit pin code to access the app. This information was not shared with HMA and the user profile as assigned to the email address used to sign up. A privacy policy was agreed between the University of Leeds and HMA to ensure compliance with GDPR regulations. Including the storage of all data on UK servers and explanations on the NON-STOP app for users to view how their data was used and their data access rights. All consent to use the app was collected by the researcher as part of the app-testing study which is described in Chapter six.

Technical support for the purpose of the app-testing study was provided by HMA as part of the contract. There were specific instances, such as issues with installing or loading the app, which were highlighted as reasons to contact HMA. To minimise participant data being passed to HMA, these were directed via the researcher to HMA support staff. This was realistic in the scope of the small-scale testing study for this project. In the future, any technical support would be received by HMA directly from the app to HMA with no input from the researcher. To provide information to the users of the app relating to things like data usage and what to do if they have issues, the NON-STOP app users had the ability to view a privacy policy as part of the download process. They were also given the option to report problems with the app. A contact email was created for users to contact HMA directly for app-usage issues.

Clinical safety in the context of the NON-STOP app was managed using two main channels of communication. If there were any concerns relating to the use of the app, the users had contact with HMA as described above. There were limited specific clinical concerns that posed a risk to the user, one example encountered during alpha testing that was highlighted was pain as a result of completing the exercises. At the intervention development stage, this was discussed and agreed that a plan would be

put in place for the app-testing study (beta-testing). This is outlined in more detail in Chapter six.

Longer-term management of the app, including any potential implementation will take place after this programme of work. To cover any 'cross-over' period between completion of this doctoral programme of work, funding was secured for intermediate hosting of the app by HMA which included 'soft' software updates. These include changes to the coding for the app to ensure it continues to work with the operating systems. It does not include any update to content. The contract also included HMA maintaining an ability to manage the app during the course of data collection and shortly after (whilst hosted) and to provide a 'kill-switch' which allows the app to be disabled entirely.

5.9 Discussion

The NON-STOP app was designed flexibly, with users in mind. This was done using a combined BCW/BCT approach, with direction from the NICE ESF and the MRC Framework for developing a complex intervention. A limitation of the BCW approach for design is that it does not formally incorporate the user-influence of designing and developing an intervention. Using it as an influence in the intervention development process, did however allow the researcher to evaluate the behavioural elements of the NON-STOP app in more detail. An extensive mapping of the MRC framework or the BCW for the NON-STOP was considered. The decision to opt against this was based on practical considerations such as time and resources, and the specific needs of the intervention. By selective mapping elements from each approach, the researcher was able to focus on the most impactful components. This approach allowed for flexibility and adaptability, making it easier to incorporate user feedback and clinical insights without the constraints and complexities of MRC Framework or BCW processes. Considering all of this, it was then possible to focus on creating a NON-STOP app aiming to maximise a change in the behaviour for children with Perthes' Disease. There are disadvantages that must be considered in not having

completed a full mapping. The key consideration being that the NON-STOP app may have overlooked important behaviour change techniques or components that could have enhanced user outcomes. However this risk was assessed in the app-testing study. Particularly in the nested qualitative study which explored users' reasons for app use.

Overall, the combined method fits with the pragmatic approach of the overarching project. The steps taken in the design and development of the NON-STOP app were based on the needs of the users and the findings of the studies completed in the earlier stages of this doctoral programme of work.

A limitation in the development of the NON-STOP app was a lack of robust, high-quality evidence to direct the content of the NON-STOP app. At the point of app-development, no randomised clinical trials have been completed to optimise non-surgical treatment of Perthes' Disease. However the work carried out in this doctoral programme, particularly the clinical consensus recommendations study [219], have been effective in providing clinical guidance in the absence of robust evidence.

Another consideration in the development of the NON-STOP app related to accessibility. The app-testing study includes usability and acceptability components that assess certain elements of accessibility. However, there are some considerations, such as digital access and literacy and online safety that it is important to clarify were considered in the development of the NON-STOP app. In the work leading to this doctoral programme, digital access was considered. Namely to ascertain what level of access to smart devices and internet/mobile data children with Perthes' Disease and their families will have. Particularly given the socioeconomic gradient associated with Perthes' Disease [14]. Whilst there are no studies assessing this specifically, the researcher was able to identify literature that provided an insight into what access is likely to look like. PPI work that took place as part of this programme of work included discussions around intervention delivery. Young children both with and without Perthes' Disease, their families, and clinicians in this patient population all agreed that an app was a reasonable and preferable mode of delivering an intervention. There were no barriers identified in this work, nor in the qualitative interview study

in this project [155] towards a digital intervention relating to accessibility. There has been work carried out by UNICEF regarding digital inclusivity for children in the UK that supports this notion, outlining the positive impact that the COVID-19 pandemic had on digital access [252]. Access to smart devices within UK households was also assessed during the 2022 census and the rate of households that did not have access to a smart device connected to the internet was less than 1% [253]. Digital accessibility is wider than access to a digital device and includes the ability of users to afford internet services to support ap use. Whilst the NON-STOP app required internet access to download it, after this, it could be used 'offline', and the progress would update once connected to the internet again. Taking all of the digital access into consideration, the researcher, as well as the supervisory team, felt confident that an app was suitable to test in the next chapter. However in a larger-scale clinical trial, a pilot phase would be advisable to monitor recruitment and any exclusion or participants refusing to take part.

Digital literacy was also considered during the development of the NON-STOP app. Various points regarding this have been made throughout the chapter. To summarise, the key points for consideration were based on the instructions given for exercises, and the information in the Learning section. Both of these were displayed in text form, and were written in collaboration with the PPI/PAG members during development. The content was written and shared with both groups and commented on. The main points for discussion and review were how understandable and readable the information was based on the age and ability of the user [254]. It was important to make sure the information met the needs of a typical child with Perthes' Disease, who is typically between four and nine years old [25]. The final iteration of the app content was shared with the app developers to be integrated to the NON-STOP app. It was outside of the scope of this programme of work to consider alternative modes of delivery for the app content such videos with as accessibility aids like voice over and captions. As outlined above, elements of accessibility were assessed in the app-testing study in the next chapter.

Online safety was not identified as a risk in the NON-STOP app development due to the app being single-user and not having any integrated components such as

communication or outside agencies such as advertising. External links that were used in the NON-STOP were selected based on prior use by the researcher and the wider supervisory team. These included links to the STEPS charity website [244] where a Perthes' Disease parents' guide was hosted. This was written by the researcher and one of the supervisory team (DP) so was deemed acceptable. Similarly, a link to a webinar, that was hosted again by STEPS charity and included the researcher and supervisor (DP). The remaining external links were governmental organisations (NHS website and GOV.uk website) and deemed appropriately safe.

In 2023, Jang and Ko outlined a framework for optimising the online safety of children and youth in Australia, Canada and the UK [255]. Whilst this piece of work was not consulted during the development of the NON-STOP app. It was reviewed by the researcher and the authors of the paper identify four risk categories. They are content risks, conduct risks, contact risks and consumer risks. The examples given in this typology of risks can be mapped to the NON-STOP app and used to demonstrate the potential risk, and the evidence of low/no risk in the NON-STOP app. These are summarised below in Table 5.8.

Table 5.8 – Online safety risks for children in relation to the NON-STOP app*

Risk category	Example of risk in practice	Risk in the NON-STOP app
Content risk	Harmful, hateful or illegal content. Or content that includes inaccurate information.	All content controlled as described in this chapter. No outside information was used.
Conduct risk	Harmful, hateful or illegal behaviour. Or user-generated problematic behaviour	No user-to-user or clinician to user communication possible within the NON-STOP app to allow this to take place.
Contact risk	Harmful, hateful or illegal/problematic encounters	No communication possible within the NON-STOP app to allow this to take place.
Consumer risk	Marketing risk, commercial profiling risk, financial risk or security risk	There was no opportunity for advertising/marketing to take place on the app. The app was freely available to users. A privacy policy which included security was agreed upon as part of the development.

The key strength of the app-development work is certainly the involvement of users in the design and the ability to implement the experiences and thoughts of key stakeholders in this novel clinical intervention. The qualitative study allowed children with Perthes' Disease, their families and the clinicians who care for them to contribute to the design and development of the NON-STOP app in a meaningful way [155]. Concepts such as avatar creation and customisation as well as the suggestion of education and information provision are perfect examples of how the intervention was developed with influence from previous findings. Having expert input from app developers with experience of creating child-healthcare apps was also paramount. The researcher provided clinical content and functionality however a collaborative approach led to the ultimate production of the NON-STOP app.

The clinical implications from the NON-STOP app at this stage are strictly limited to the next stage of the project. Completion of the development allowed progression to Chapter six, which was to undertake some introductory testing of the NON-STOP app prior to a larger-scale clinical trial.

5.10 Conclusion

This chapter summarised the approach taken to design and develop the NON-STOP app, a digital self-management intervention for the non-surgical treatment of Perthes' Disease. A pragmatic approach, with input and guidance from experts as well as relevant methodologies and theoretical underpinning has led to the creation of a novel intervention suitable for further testing in a small-scale study. The usability and acceptability of the NON-STOP app is described in the next chapter.

Chapter 6 – Testing the usability and acceptability of the NON-STOP app

6.1 Introduction

The chapter describes the final study, which aimed to test the practical application and use of the digital self-management intervention, the NON-STOP app. At the time of submission this paper has been submitted for publication and is under review.

The links to the theoretical underpinning of the study design are described within the context of the overarching theoretical approach taken in the programme of work. A mixed method design was employed, included both quantitative and qualitative components. The quantitative component of the study explored app usability and acceptability whilst the qualitative component explored users' experiences of the NON-STOP app. Finally the chapter summarises the findings in relation to what is already known, what this study adds and the clinical implications.

In reference to the MRC framework, this study maps to the "Feasibility" stage of the framework (shown in Figure 2.5). The core elements of relevance at this stage were engaging stakeholders and to refine and retest the programme theory from the Logic Model (Appendix I). It is worth noting that this study was not a true feasibility study, rather that it describes the element of the MRC framework that it maps to. More detail on this is provided in Section 6.3.

6.2 Aims and objectives

Aim

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

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.

6.3 Study design

A mixed-methods design was employed in this usability and acceptability study. More detail regarding the rationale for the methodological approach taken has been presented in Chapter two. The quantitative component, hereafter referred to as the 'before and after observational study', was an observational study in which participants used the NON-STOP app for six weeks [256]. App use was monitored using data directly from the app and participants completed PROMs before and after app-use. As part of the quantitative component, there were feasibility objectives to assess some study processes such as pre- and post-app testing outcome collection and participant retention. Some uncertainties would still need to be addressed in an internal pilot phase of a trial, but were useful here to inform planning. These are discussed in more detail in this section.

The qualitative component, hereafter referred to as the 'nested focus group', was completed after completion of the before and after observational study [257]. This focus group involved some participants from the before and after observational study. During the focus group, the experiences of app use were explored in more detail as well as opinions on any changes needed to improve the app.

6.3.1 Theoretical underpinning

This final study of the doctoral programme of work continues to be underpinned by the same theoretical and philosophical approach i.e. pragmatism. Similarly, the behavioural theories of SDT and the SEM were also applied which have been presented in more detail in Chapter two [105].

SDT provides an explanation for what influences individuals to adopt and maintain new behaviours [78]. Using mixed-methods, the researcher assessed the impact that the intervention had on self-management behaviours. The NON-STOP app was designed to be fun, provide rewards and be relatable to the user (a child with Perthes' Disease). The intent, based on SDT, is to increase physical activity and completion of exercise programmes underpinned by an increase in participant autonomy, competence and relatedness. This was assessed using pre- and post-app use questionnaires as well as monitoring activity through the app. In the nested focus group, questions were asked related to these behaviours.

SEM places more of a focus on the environmental factors that the behaviour takes place in [109]. Whilst more of a contributing theory in this app-testing study, it was a key component of adapting the environment in which these behaviours took place. Making a digital intervention that was accessible everywhere was important to meeting the environment needs of the user. The hypothesis in the study was that making the NON-STOP app accessible to users, would lead to an increase in use. Questions in the nested focus group related to this theory were based on where/when a child might have used the NON-STOP app and how this was different to their previous self-management.

Epistemological stance

The final study adopted a pragmatic epistemological stance, consistent with the wider programme of work [42]**.** This mixed-methods study aligns well with the pragmatic paradigm. In order to test the usability and acceptability of a digital self-management intervention for Perthes' Disease, it was reasonable to have children with the

condition, and their families test it for a short period of time. Similarly, it was reasonable to suggest that using the NON-STOP and only capturing quantitative data app would not give rich, meaningful information about how the app functions and fits into the life of a child with Perthes' Disease and their family. The pragmatist links knowledge to experience [258]. With this in mind, focus groups were planned for after the app-testing period. Along with support from the supervisory team, these methods were deemed optimal to meet the study aims and objectives.

6.4 Sample and setting

The study eligibility criteria and recruitment procedures were similar to the qualitative study in Chapter three. Participants were children with Perthes' Disease, receiving treatment in one of three NHS hospitals (Leeds Children's Hospital, Alder Hey Children's Hospital and Sunderland Royal Hospital). Recruitment from different centres was important to ensure the sample is representative of the wider UK patient population. Participants were recruited from their usual orthopaedic appointments in which they are regularly assessed by an orthopaedic consultant. Patients were sampled purposively to maximise heterogeneity e.g. differing sex, age, treatment exposure, duration of living with condition and disease severity. The children recruited were initially identified by a lead-clinician within each centre who was familiar with the study eligibility criteria.

For the nested focus group, a sub-set of the existing sample were invited to take part. After providing trial consent, participants were asked to complete an online questionnaire to provide some demographic and disease-specific information. At this point they were asked to provide a response to whether or not they were interested in hearing more about taking part in a focus group in the future. This was done with a simple "yes" or "no" response on the online survey.

6.4.1 Eligibility criteria

Participants (including family members) were eligible for inclusion if the child:

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

Participants (including family members) were excluded if:

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

The rationale for excluding children with Perthes' Disease who had recently undergone surgery within the previous six weeks was that children were required to complete physical activity as part of using the app. Typically, children are placed in casts post-operatively to maintain ROM, or are given weightbearing restrictions and would not have been able to complete the exercises/activities advised within the NON-STOP app.

6.4.2 Sample size

The sample size selected for each of the components in this study was based on a realistic, achievable number that would effectively address the aims and objectives. It varied within the two components, where there were specific considerations.

6.4.2.1 Before and after observational study

A sample size of 30 children was selected for the before and after observational study. Sample size calculations for before and after observational studies are not well described in the literature [259]. Often sample sizes are determined based on the number of potential participants available [260] combined with aims and objectives typically relating testing how the intervention rather than clinical effectiveness based on power. To test the NON-STOP app, the sample size was deemed adequate given that the study objectives, which was to examine app acceptability and usability rather

than clinical effectiveness. This is supported by the work of Eldridge et al work, which demonstrated similar studies that assessed feasibility of an intervention in children [152]. The sample size was also thought to be achievable given the previous experience of recruitment by the researcher in the earlier stages of this project.

6.4.2.2 Nested focus group

In qualitative research, the aim is to produce data that is meaningful and allows the researcher(s) to best understand the phenomenon in question [261]. In this nested focus group component, the sample size was selected at up to five child/family pairs, hereafter referred to as 'dyads', from each recruiting site. Totalling three focus groups. In a literature review by Adler et al, the authors summarised aspects of focus group study design for child, youth and parent research [262]. In their review, the authors described a number of research studies that recommended a focus group size of between 3-10 children. In the nested focus group of this study, a sample size of up to five dyads allows for a small enough group for children to benefit from a peer-audience, but also not feel overwhelmed by the number of participants.

6.5 Recruitment

The recruitment processes for each component of the feasibility study are described below.

6.5.1 Before and after observational study

Children with Perthes' Disease were recruited from their existing clinical appointments. This took place in three centres within the UK. At Leeds Children's Hospital, the researcher was the clinician that recruited participants. At the two remaining sites (Alder Hey Children's Hospital and Sunderland Royal Hospital), an appropriate clinician (site PI) at each centre was identified. The clinicians were consultant orthopaedic surgeons with a specialist interest in hip conditions including

Perthes' Disease. Each site aimed to recruit ten children with Perthes' Disease to achieve the overall target of 30 participants.

At the clinic appointment, any child that met the eligibility criteria had the study briefly outlined to them and if interested, were asked permission to share their contact details (name and email address) with the researcher. These details were sent by the clinician to the researcher using a secure email domain (NHS.net). As in Chapter three, agreement to pass on contact details did not constitute consent to taking part in the study. The permission to contact the family member was recorded in the patient's medical notes by the recruiting clinician. The researcher then contacted the potential participants with the participant information sheet (PIS) for child, shown in Appendix O and adult, shown in Appendix P as well as instructions on how to provide consent. The overall process for recruitment to the before and after observational study is illustrated in Figure 6.1.

6.5.2 Nested focus group

Participants who took part in the before and after observational study were invited at the start of data collection to indicate whether they were willing to receive more information regarding the nested focus group. Those who expressed an interest were emailed two weeks before the end of the app-testing component, and invited to give consent to take part in a focus group. The email provided a PIS for child Appendix Q and adult Appendix R, outlining the purpose of the study and included instructions on how to provide consent.

Participants who replied providing valid consent were then invited to take part in a focus group at their local clinical centre. It was clear in the invitation that the focus groups were to involve the child with Perthes' Disease that had used the NON-STOP app and a family member (a dyad). The aim for the nested focus group was to recruit up to five dyads from the ten recruited children at each site. More detail on the process of the nested focus group can be seen in Figure 6.2.

6.6 Consent/assent

Informed consent was gained for all participants. In the before and after observational study, this was provided by a parent/legal guardian of the child with Perthes' Disease. In the focus group, this was once again provided by the parent/legal guardian both for themselves as well as the child. In addition, children were asked to provide assent to participate. The guidance that surrounds it, has been discussed in Chapter three.

6.6.1 Before and after observational study

After an appropriate child with Perthes' Disease had been identified and approached, the parent/legal guardian provided an email address. An email was then sent by the researcher to the parent/legal guardian, outlining their involvement in the trial, and clear instructions for how to provide consent to take part in the research study.

As with the approach used in the first study of this programme of work, a proportionate consent process was appropriate given the low-risk nature of the data collected [174]. The parent/legal guardian was asked to consider the information provided, and then to reply to the email and include the 'statement of agreement' in their reply. The statement was copied from the email and returned to the researcher. It read as follows:

> *'I have studied the information provided in the participant information pack and understand what will be required of me/my child during this study. I consent to participate in the study to test the NON-STOP app. 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 participation is voluntary, and I am free to withdraw from this study, unchallenged, at any time.'*

The protocol included a plan for obtaining written consent in cases where potential participants did not have access to e-mail, however this did not occur. If this had

arisen, a copy of the statement of agreement would have been printed and sent by post to the participant.

After returning the email and statement of agreement to the researcher, the participant (child) was progressed to the next stage of the before and after observational study. This information is outlined in the data collection section below and can also be seen in Figure 6.1.

6.6.2 Nested focus group

As outlined in the recruitment section above, participants (child/family dyad) who consented to the before and after observational study were asked to complete a pre-testing questionnaire. In this questionnaire, there was an option to be contacted at the end of the study regarding a focus group study that was planned to further explore the usability and acceptability of the NON-STOP app. The answers were "yes" or "no". Participants who selected yes, were contacted at the end of the six-week period and invited to take part in a focus group in their local area to discuss the use of the app. The invitation to take part was sent via email, with the PIS and instructions of how to provide consent to take part in the focus group. This approach was almost identical to the consent process for the before and after observational study.

The parent/legal guardian of the child with Perthes' Disease was instructed to read the information (PIS for both family and child participant) and reply with the 'statement of agreement'. This had been adapted for focus group participation and read as follows:

> *'I have studied the information provided in the participant information pack and understand what will be required of me/my child during this study. I consent to take part in the focus group study regarding the NON-STOP app. 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 participation is voluntary, and I am free to withdraw from this study, unchallenged, at any time.'*

After returning the email to the researcher with this statement, the next step of the nested focus group took place. More detail on the nested focus group is provided in the data collection section below and can also be seen in Figure 6.2.

At the focus group, participants (dyads) were once again asked to provide their verbal agreement to take part in the study. Assent was gained from the children who took part in the study. Assent is a concept that is not straightforward, and has been considered and discussed in this thesis in more detail already in Chapter three. If a child were to have verbally refused to assent to the focus group study, they would have not taken part, and the parent/legal guardian would still have been offered the opportunity to participate. This did not happen in the study. There were confidentiality considerations for the nested focus group which are discussed in the ethical considerations section below. However it is important to note that for the purpose of the consent process, all participants were informed that no information shared in the nested focus groups would be shared with their treating clinical team.

Figure 6.1 – Before and after observational study process

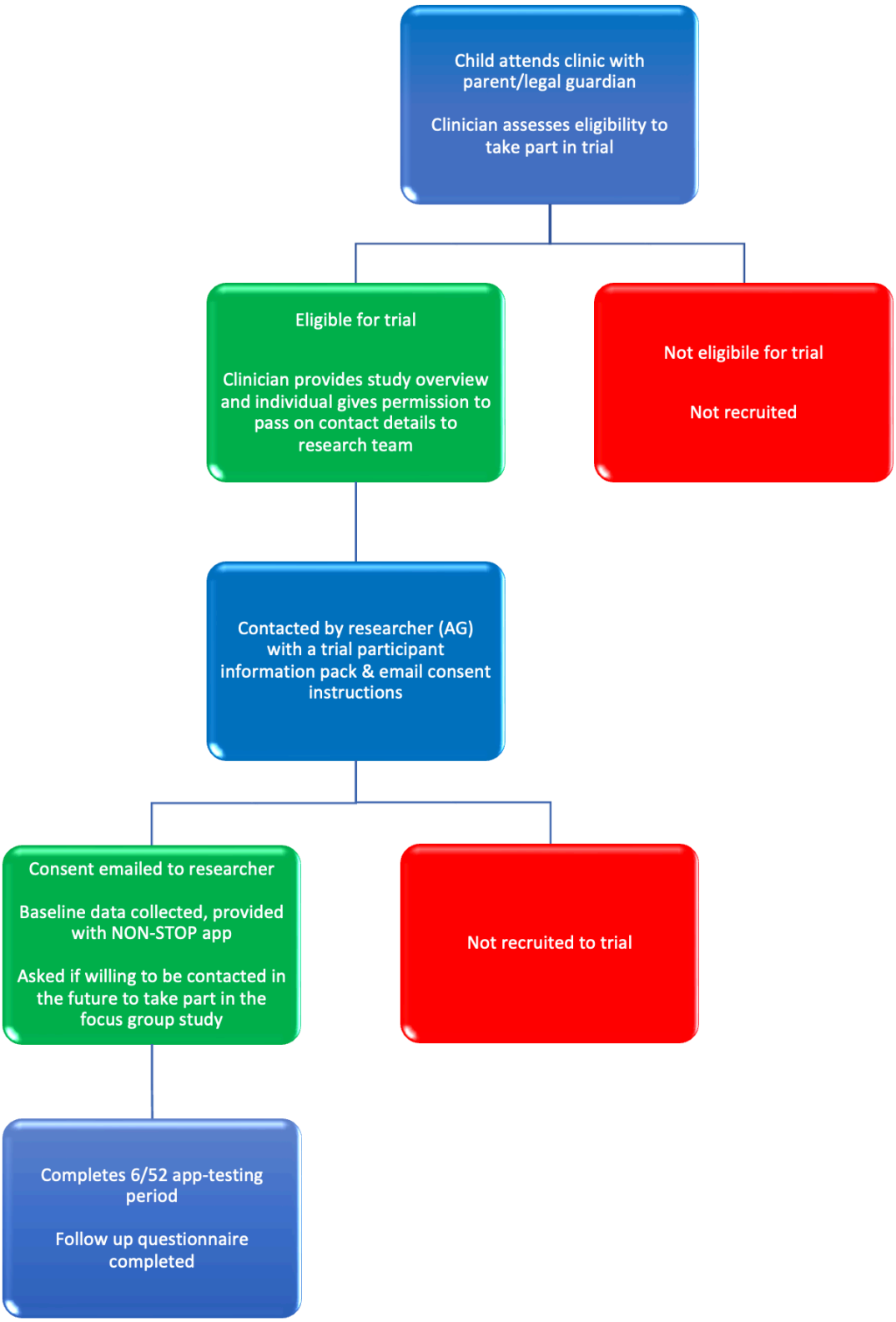
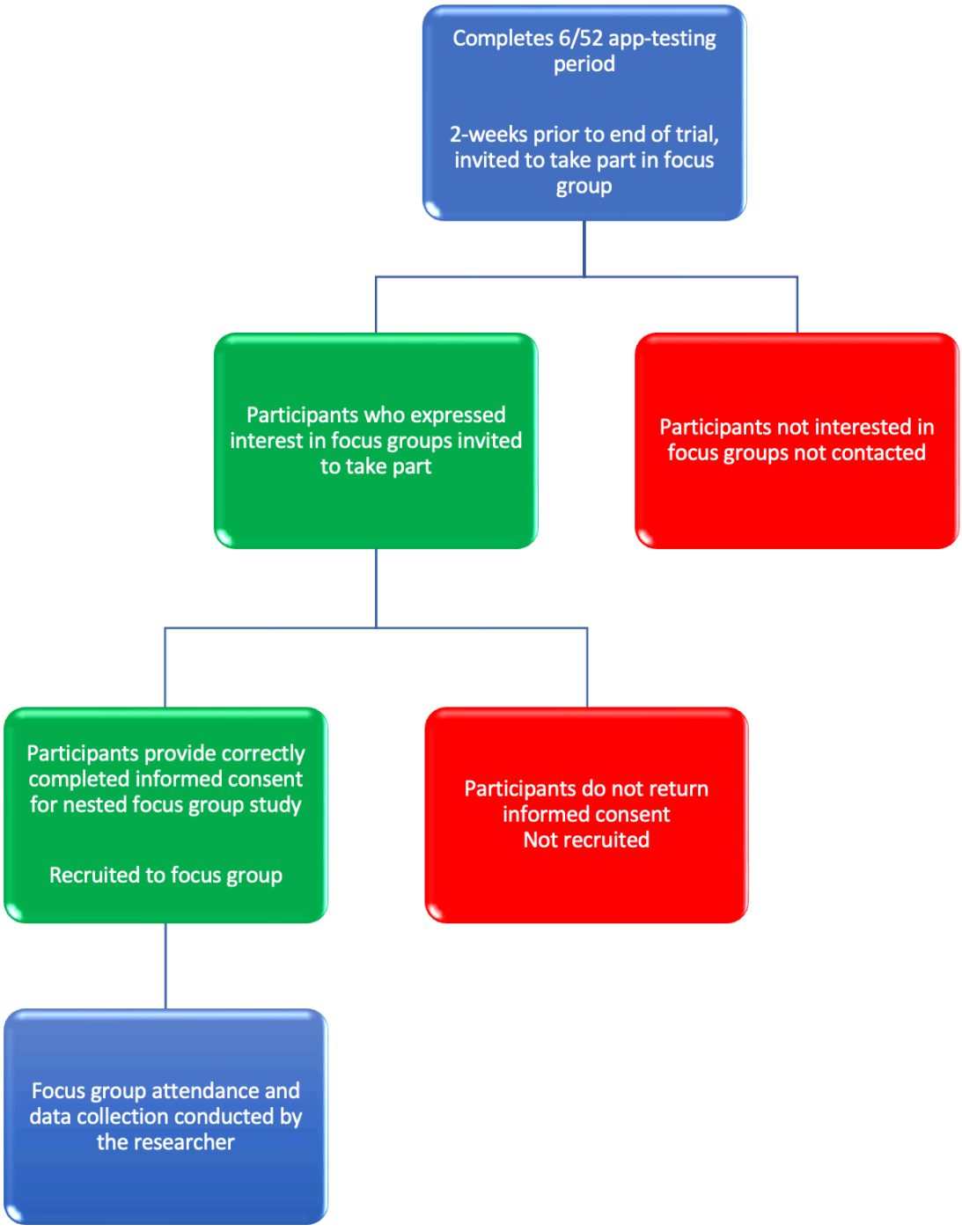


Figure 6.2 – Nested focus group process



6.7 Data collection

The data collection methods for the two elements of the usability and acceptability study are provided below.

Demographic information

Demographic information was gathered to describe the patient population and to explore the representativeness of the trial population compared to the wider clinical population. Demographic information collected included:

- Age at diagnosis (years)
- Age at time of recruitment (years)
- Sex (male/female)
- Ethnicity (White, Mixed/Multiple ethnic groups, Asian/Asian British, Black/African/Caribbean/Black British, Other ethnic group (free-text box option provided if selected)).

The epidemiology of Perthes' Disease has been well described [25, 263] and it was important to, where possible, try and replicate the rates of these characteristics within the study sample. As discussed in the results section below, this is reflected with higher proportion of males to females in the sample, however the rates of female representation are at least 20% which is in line with the results seen in the BOSS study [25]. Similarly, it was possible with purposive sampling to ensure that patients from a varied ethnic background were also approached, following national guidance for collecting ethnic data in research [264]. The data were collected in an attempt to ensure that the views and experiences of as many as possible were gathered.

Recruitment data were also collected from each site. Including the total number of potential participants invited, the number that consented to the trial and finally the number of participants that completed the study.

6.7.1 Before and after observational study

Data collected in the quantitative arm of the study reflected that of the CONSORT Extension for Pilot and Feasibility studies to guide transparent reporting of study processes and outcomes [265]. This is of particular use when preparing for a more definitive clinical trial, as was the case. Appendix S shows the CONSORT checklist, note that some elements are not completed, this is because the study was not a standalone pilot or feasibility study, as discussed earlier in the chapter.

Trial participants were required to provide data at two time points, initially before they were given access to the NON-STOP app for testing and the second after the six-week testing period. A summary of the scheduled assessments is provided below in Table 6.1.

Table 6.1 – Content and timing of assessments during before and after observational study

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

Baseline data included participant demographics and patient reported outcomes. Participants were given a link to provide the baseline data in an online questionnaire, hosted using Online Surveys [193]. The outcomes including the Patient Reported Outcomes Measurement Information System (PROMIS) Mobility [103] and Children's Physical Activity Questionnaire (CPAQ) [266, 267]. Outcome measures were collected at appropriate times during the study period. For instance, Health ITUES was not collected until after testing the NON-STOP app as it relates to usability of an intervention.

Each measure is discussed in more detail below. However, the aim of this study was to test methodological uncertainties such as data completion, recruitment and the usability and acceptability of the NON-STOP app. This meant it was more important to gather the number of responses than it was to study the content of the responses.

To give an example, in this before and after observational study, the change in score PROMIS Mobility at each time point was not the study outcome. The outcome was whether the participant completed the outcome measure before and after the app-testing period. Testing completion provided a surrogate measure of feasibility regarding outcome data completion, which is useful for any study in the future using these outcomes.

At the end of the six-week period, participants completed the same outcome measures as pre-testing (PROMIS Mobility and CPAQ). An additional outcome measure was also completed, the Health Information Technology Usability Evaluation Scale (Health-ITUES) [268].

Patient Reported Outcomes Measurement Information System (PROMIS) Mobility

The PROMIS Mobility score is a validated patient-reported outcome measure for children with Perthes' Disease [103]. More detail about the outcome measure is discussed in Chapter two where PROMs relevant to children with Perthes' Disease are discussed. In 2020, a core outcome set was developed for Perthes' Disease, and the PROMIS Mobility was included in this [95]. The inclusion of PROMIS Mobility within this core outcome set was a strong influence on its selection for this before and after observational study. It is a self-, or proxy-reported measure of physical function. 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 [240, 269]. The use of the PROMIS Mobility outcome in this thesis is in line with the guidance from PROMIS Health Measures group regarding obtaining and administering measures [270]. Those wishing to use PROMIS measures should review the host website and review the guidance before using the tool.

The most up to date version of the PROMIS Mobility is provided in Appendix A. PROMIS Mobility asks users to provide a response of how easy they find activities of daily living. A Likert scale is used, ranging from "With no trouble", which scores four points, to "Not able to do" which scores one point. The tasks range from simple tasks

such as “I could walk across the room” to more advanced activities like “I could run a mile”.

PROMIS Mobility is then analysed by converting the total raw score of the measure into a T-score. This conversion allows the user to compare scores with the “general population mean” which was produced as part of the development of the PROMIS Physical Function tools (of which PROMIS Mobility is a subset) [103]. A higher score indicates more of the concept being measured, i.e. better mobility and a higher level of function for individuals [269]. In this before and after observational study, participants were asked to complete this score at the start of the trial then again at the end of the six-week testing period.

The main aim of using this outcome measure as part of the feasibility study was to assess whether an adequate number of participants completed their follow-up assessments after testing the app. The more granular data from the scores could be analysed in more detail, however it is unlikely that a six-week app testing period is suitable to detect any statistically (or clinically) significant difference in each child with Perthes’ Disease.

Children’s Physical Activity Questionnaire (CPAQ)

CPAQ is a parent-reported 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 [267, 271]. There are no studies involving children with Perthes’ Disease where CPAQ has been used. However, in a study by Corder et al, the CPAQ was compared with four other self-reported physical activity questionnaires [267]. CPAQ was deemed to have moderate levels of reliability and usability when compared with others, and whilst others were recommended for ages 12-16 years old, CPAQ was more appropriate for much younger children.

CPAQ asks whether the child had completed activities over the last seven days. The list includes commonly completed sports such as football, gymnastics and swimming, as well as leisure activities like playing on playground equipment or taking part in

Physical Education at school. CPAQ is reported in frequency of activity completed, and minutes per day for each activity [271]. In the online questionnaire, participants were asked to select “yes” or “no” to a range of commonly completed exercises and activities that they may have done in the last week. If they selected yes, they were asked to provide the number of minutes they had completed the exercise. The most up to date version of CPAQ is provided as Appendix T**.** Participants were asked to complete this tool before they started using the NON-STOP app, and then once again after they had tested the app for six weeks.

It is possible to carry out inferential statistics using CPAQ results. Corder et al described methods of outcome derivation based on the energy expenditure of common activities [267]. This was done using an extrapolation of published data for older children [272] and would have been possible to use this data to calculate a Physical Activity Energy Expenditure value that could have been assessed pre- and post-app testing. This is something that would be considered for a larger-scale trial where clinical effectiveness was to be measured.

Health Information Technology Usability Evaluation Scale (Health-ITUES)

Health-ITUES is a validated measure used to assess usability of a digital tool [273, 274]. This 20-question, customisable questionnaire is used to assess usability and acceptability in four domains [275]. The four domains are impact, perceived usefulness, perceived ease of use, and user control. All of which related 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. The Health-ITUES for this study to test the NON-STOP app was included in the post-trial information. It can be seen in as Appendix U.

The participants who had tested the NON-STOP app were sent a link to complete this information using an online survey as described. The data from this provided an average score in each of the four domains:

1. Impact
2. Perceived usefulness
3. Perceived ease of use
4. User control

The scores in each domain are given a rating of 1 to 5. An average (mean) score for the cohort is then calculated for each domain, and for the whole questionnaire. Previous research has identified an optimal usability cut-off score of 4.32 [276]. This was not completed in the same patient population, and no research has been completed for physical interventions usability for children with Perthes' Disease. In the absence of this, it was deemed appropriate to use this score to look at the intervention in each domain. For example, to calculate the mean score in the 'Perceived ease of use' domain, then check against the usability cut-off score, and then consider whether it could be deemed easy to use or not. Similarly, an overall score within the four domains can be calculated by taking the mean of all 20 items on the Health ITUES.

Process measures

The before and after observational study also included gathering process measures from the app. Process measures in the context of digital technologies refer to the measures of real-time data that are used to report use [277]. These were collected as part of the participants' use of the NON-STOP app and gave an insight into how active participants were, and what parts/aspects of the NON-STOP app were used most. This information was used to influence the nested focus group discussion. The direct influence of the app data is discussed in the nested focus group section below.

The measures were selected using the pragmatic approach that underpins this project. However, the Template for Intervention Description and Replication (TIDieR) checklist was used as a method of ensuring important components of an intervention were considered [278]. This checklist was created by Hoffman et al in 2014 with the aim of improving how interventions are reported in studies [279]. Typically, this is reported by the authors who developed the intervention in the protocol. It is then

possible for readers, reviewers and editors to understand the intervention in the necessary detail [279]. In this study, the TIDieR checklist was used to select the process measures. For instance, the frequency and duration of app use, and the specific activities completed by the participants.

The process measure information was collected directly from the NON-STOP app by the app-development company, unidentifiable information was collected using each participant's unique identification number. After downloading the app, they completed the "My Journey" training package and then completed at least one activity session, hereafter referred to as "onboarding".

The process data that was collected was:

Initial engagement:

- Number of invited participants that registered
- Number of participants that completed the training package (worked through the "My Journey"). i.e. "onboarding"

NON-STOP app use (for those that completed onboarding):

- Number of app log ins
- App log ins per week
- Number of times met target for progress reward
- Average pain score (average based on daily score from individual)
- Highest/lowest pain score (range based on daily score from individual)
- Most popular activity used

Pain was measured using the Wong-Baker FACES Pain Rating Scale, seen in Figure 6.3 [236, 280]. App users provided their pain score once per log-in and there were clear instructions advising them to contact their existing clinical care team if they had specific questions or concerns about their pain levels. The pain score that they selected dictated how long they were advised to complete their chosen activities for.

Figure 6.3 – Wong-Baker FACES scale*

**Wong-Baker scale adapted from Garra, G., et al., Validation of the Wong-Baker FACES Pain Rating Scale in paediatric emergency department patients. Acad. Emerg. Med, 2010. 17(1): p. 50-4.*

6.7.2 Nested focus group

Separate focus groups were arranged in each of the three recruiting sites of Leeds Children's Hospital, Alder Hey Children's Hospital and Sunderland Royal Hospital. As with the before and after observational study, number of participants/dyads approached and consented were collected and are provided in the results section below. The focus groups took place in a neutral space identified at each of the sites. A non-clinical environment was used in order to facilitate an open, honest discussion between participants and the researcher. Communication with Alder Hey took place using existing relationships with the extended working team of project supervisor (DP) who also acted as recruiting clinician for Alder Hey. In Sunderland, the recruiting clinician also facilitated the booking of a youth team space that was suitable for use of up to 30 people.

The focus groups were conducted by the researcher, and an independent facilitator supported by providing field notes from the focus group. Focus groups were recorded using an encrypted audio recording device. The recordings were then exported to secure University of Leeds servers in password protected files. They were then deleted from the audio recording device. The recordings were sent to a University-preferred supplier for transcription, as used in the qualitative study in Chapter three.

The independent facilitator (not the researcher) at each site was a children's specialist physiotherapist with experience of communicating with children with Perthes' Disease and their families. They assisted in facilitation of discussion within the focus group, as well as taking field notes to accompany transcripts. This was done to supplement the recordings and mitigate for any failures in recording, but also to provide valuable context to conversations where audio recordings may not capture. It would have been possible to video record the focus group. Video recordings have been used more in focus group work recently, particularly in the video-conferencing space [281]. Video analysis can provide insight into things like body language, hand gestures and facial expressions. By contrast, inclusion of video recording equipment may also have been intimidating to participants and reduced engagement. A single recording device, and someone taking notes does pose a similar risk, but an additional camera set up may compound this. There are also considerable costs and resource-needs to consider that would have been beyond the scope of this project. Field notes by an independent facilitator were therefore deemed appropriate, and are recommended for documenting context in qualitative work [282].

The focus groups followed a topic guide, shown in Appendix V, which was developed using a combination of sources. There was influence from the theoretical underpinning of the project, namely SDT which considers autonomy, competency and relatedness. The topic guide for the focus groups was reviewed by the Project Advisory Group and a local NIHR Young Persons Advisory Group. At the focus groups, the researcher used the topic guide to explain the study, deliver information relating to the ground rules seen in the topic guide. Guidance on confidentiality within and outside of the focus group was stressed to encourage engagement with the focus group. After this, a final check of consent/assent with participants was conducted, recording began, and the focus group took place.

Questions/topics considered the self-management elements of the NON-STOP app and whether the functions of the app met the needs of the users. The topic guide was also influenced by the results of the before and after observational study. Reviewing the data from the app-testing element of this study allowed the researcher to prompt participants and ask questions relating to specific parts of the app. To give

an example, questions regarding the activity section were tailored to understand why the most popular activity was such, and vice versa. Similarly, getting an insight from those that used the NON-STOP as to why parts of the learning section were not accessed. The before and after observational study allowed the researcher to identify pages that were viewed a small number of times over the six-week testing period. The nested focus group gave an insight in to why these may have only been used such a small number of times.

More information on data analysis from the focus group is provided in section 6.8 however it is important to note that the first of the three focus groups acted as a 'pilot' for the remaining two. After the first focus group, the researcher carried out a reflection, considered the topic guide and what prompts worked well/not so well. These were then adapted accordingly for focus groups two and three. As with the quantitative component, the qualitative component was informed by reporting guidance. In this instance, the Consolidated Criteria for Reporting Qualitative Research (COREQ) was used to ensure transparency in methods and acknowledgement of reflexivity [283]. The COREQ checklist is attached as Appendix W.

6.8 Data analysis

The analytic approaches for the mixed-methods study have been separated into the two component studies. A summary of the impact that the before and after observational study data had on the focus group is provided in the results section.

6.8.1 Before and after observational study

Baseline characteristics

Participant characteristics were reported to ensure that the participants in the trial were representative of the Perthes' Disease patient population and in line with guidance for reporting ethnic background in health research [25, 264].

Demographic information was analysed using descriptive statistics. Age data were expressed as a mean (range), whilst sex (n, % male) and ethnicity (n, % White British) were expressed as frequencies and proportions. Whilst other studies may opt for standard deviation measures when describing variation in participant age, it was deemed more appropriate to report the age range. The common age of diagnosis for Perthes' Disease is between four and nine years old [263] and reporting the age range allowed for a more meaningful clinical comparison. For sex, Perthes' Disease affects males disproportionately more than females, therefore the largest proportion has been reported. Similarly the most common ethnicity in the British population of children with Perthes' Disease has also been reported [25].

Outcome measures

Inferential statistics were not carried out as this before and after observational study was not powered to detect meaningful statistical or clinical changes in outcomes between baseline and follow-up. The descriptive analysis focused on assessing the function of the digital intervention rather than identifying any treatment effect. All outcome measures (PROMIS Mobility, CPAQ and Health ITUES) were analysed using appropriate descriptive statistics at baseline (when assessed) and 6 weeks follow-ups.

PROMIS Mobility

PROMIS Mobility scores, as discussed in the data collection section, were not analysed using the scores in the domains. The total number of respondents (also reported as proportion) were reported.

CPAQ

For the purpose of this usability and acceptability study, descriptive statistics relating to pre- and post-app testing completion of the measure were used. Description of activity levels gave an overview of whether children did more or less activity in

general after using the app could be considered however were considered to be outside of the scope of this project.

Health ITUES

In the before and after observational study, this was done for participants who completed the post-testing information. The total usability score for all 20 items as well as each domain have been reported in the results section. With the usability cut-off score of 4.32 used to demonstrate 'usability' of the intervention.

Process measures

For each of the process measures gathered, the list below outlines the descriptive analysis that took place. Frequencies counts (n, %) or a measure of central tendency and the associated variation were presented at 6 weeks. As the data were not normally distributed, the median and the inter quartile range (IQR) were presented as opposed to the mean and standard deviation (SD). This is because outliers existed within the data as users who engaged with the NON-STOP app once and then stopped, and a small number who did not engage at all. The presence of these users in the data set caused a skew in the data which meant that the IQR was the most appropriate measure to use to summarise the data [284].

Initial app engagement:

- Number of invited participants that registered (number and %)
- Number of participants that completed the training package (worked through the "My Journey") i.e. "onboarding" (number and %)

NON-STOP app use (for those that completed onboarding):

- Number of app log ins (cohort total, median (IQR) and range)
- App log ins per week (cohort total, median (IQR) and range)
- Met target for progress reward (% each week)
- Average pain score (cohort total, median (IQR) and range)

- Highest/lowest pain score (cohort total)
- Most popular activity used (cohort total)

In the results section the total values are reported as above, the results are also described. This is to provide an overview of trends and patterns of elements such as log ins within the cohort. The most visited pages within the NON-STOP app are also discussed, as are the differences in total app use for the cohort.

6.8.2 Nested focus group

The Framework Method (Framework Analysis) was used to analyse the data from the nested focus groups. This method had been used already in Chapter three. Using the Framework Method once again gave the researcher a focused approach for analysing this qualitative data by organising it into codes that aligned with the aims and objectives of the study. The structured categorisation enabled the researcher to reach a conceptual understanding of the subject under investigation, i.e. the usability and acceptability of the NON-STOP app. This structure was applied to ensure that the method could address the aims of the study whilst still allowing for the interaction of participants to be captured using emerging themes.

Transcripts of the focus groups were analysed using the stages of the Framework Method [121] used in the qualitative study in Chapter three. It follows the five steps of familiarisation, coding, indexing, charting and mapping and interpretation which are described in more detail in Chapter two.

The coding matrix was designed based on the aims and objectives of this usability and acceptability study, however it was important that it aligned with the theoretical underpinning of the project. The theoretical approach has been extensively described in this thesis in Chapter two, and summarised in the context of the NON-STOP app in Chapter five. Each code relates to a specific element of the project and how it was underpinned by theory, the study aims, and importantly, with the involvement of key stakeholders at important milestones within the doctoral programme of work. A summary of the coding matrix is presented in Table 6.2.

Table 6.2 – Coding matrix alignment with theory

Coding label	**Alignment with approach in project**
1. Usability	Specifically this coding label aligns with SDT in that it relates to "Autonomy" and "Competence" [78]. i.e. if the NON-STOP app is easy to use independently, it increases the autonomy of the user (the child) which hopefully leads to a sustained change in behaviour (sustained use of the NON-STOP app).
2. Rewards	Rewards are well described in the Behaviour Change literature [145] and have been discussed in this thesis. The rewards within the NON-STOP app were not only supported by BCW theory, but discussed in the qualitative study in this project [155].
3. Experience of the app	Identifying elements of the NON-STOP app that children/families did not like once again aligned with SDT. It specifically aligns with the "Relatedness" element of SDT. Understanding elements of the NON-STOP app that users had positive or negative experiences of is important to consider how that affects long term behaviour (app use).
4. Different to normal/previous care	Understanding how this novel digital self-management intervention fit in to children/families' routine was important. This aligns with the SEM which explores the involvement of others (described in the "Interpersonal" domain of the model) [109]. In the focus groups this coding label allowed the researcher to analyse reports of family support and self-efficacy.
5. Future use	The aims and objectives of the usability and acceptability study outline the importance of testing the intervention prior to a definitive clinical trial. Exploring future use and any potential refinement/modification was important, thus the need for this coding label.

Three focus groups took place. An iterative approach involved a review of the analytic coding framework after the first focus group. After applying the coding framework, it was checked for how applicable it was to the data. A review with supervisors with expertise in qualitative research (SR) and wider research team (SP) facilitated discussion and justification of changes made. This review process also allowed for a discussion on any emergent codes from the data collected in the focus groups. The

analytic coding framework can be seen in Table 6.3**.** There were no changes made to the coding framework as a result of analysis process; deductive coding found this matrix adequately captured main themes. However there was an emergent (inductive) code, which related to additional points raised about the app. This focused on who the app was used with (school staff, sports clubs, etc.) and how many times per week was acceptable to the users.

Table 6.3 – Analytic framework developed to support focus group coding

Coding label	Notes/ideas	Child description	Family description
1. Usability	Points around downloading, logging in, instructions/training package, reminders and time consuming	- Points around being able to use it themself. - Recall of any elements of the training package (videos, cartoons, etc.)	- Identification of child using the NON-STOP app independently. For example using a parent/guardian's phone without help. - Mention of reminders being used/helpful
2. Rewards	Child-specific reward such as avatar for completing exercises	- Child mentioning rewards that they gained, or how they customised their avatar after doing their exercises.	- Points about the reward system and how it influenced the use of the NON-STOP app
3. Experience of the app	Fun or not. What liked/disliked (with specifics)	- Any aspects of the NON-STOP app that were discussed either in positive or negative way.	- As with child description. Specificity to exercises/activities or learning section.
4. Different to normal/ previous care	Comparisons relating to time taken to complete, frequency completed on average week	- Any mention of the NON-STOP app in comparison to their existing care/routine such as physiotherapy programme or information about their Perthes' Disease.	- Points around compliance/adherence and whether child found it more/less fun and engaging.

5. Future use	Points made around future care with or without app	- Ideas for the future of the NON-STOP app such as new characters, exercises. - Changes to the layout/structure of the NON-STOP app.	- As with child description
6. Additional points for app use	Considerations around the use of the NON-STOP app	- Anything about use of the app that was not covered by the codes.	- Discussions of things such as dosage/frequency of using the app - Inclusion of other members of the psychosocial make-up such as school, clubs, etc.

As with the qualitative interview study (Chapter Three), it would have been possible to use other data analysis approaches for this study. Thematic analysis in particular could have been useful here given its focus on the experiences of the participant [125]. However the aims of this study were relatively focused on reviewing the usability and acceptability of the NON-STOP app. Specific categories for discussion are more aligned with the Framework Method rather than Thematic analysis which often starts with more emergent coding and may have been less aligned with the study objectives [126]. Similarly, content analysis can be used in qualitative research, however in the context of this study it was not chosen. The most significant factor was the depth of analysis that the method offers. Content analysis focuses on frequency and presence of specific words or phrases [123]. This approach could have been used in this study to link to things like the theoretical underpinning, for example presence of phrases like “reward”, “fun” or “independence” that would link to psychological theories within this project. However when compared to the Framework Method, it does not allow for as much of an in-depth analysis of the themes or concepts discussed.

Data were stored using NVivo software (Version 12, March 2020) [175] where the coding framework was applied.

6.9 Ethical considerations

NHS ethics and Health Research Authority (HRA) approval was obtained given the involvement of NHS patients. This study was submitted as a whole (before and after observational study and nested focus groups) to NHS Research Ethics Committee (REC) and HRA. It was awarded a favourable opinion by NHS West Midlands – Edgbaston REC 22/11/2023. REC reference 23/WM/0251, the letter outlining the favourable opinion is attached (Appendix X)**.** Sponsorship was provided by the University of Leeds and local research management and governance approval for each NHS trust was obtained from their research and development departments.

Ethical considerations such as involvement of children in research and subsequent deliberations over consent/assent processes were discussed in Chapter three and were followed in the same way for this final study.

Consideration of participant time was once again a factor. However through work completed as part of this overarching programme of work including qualitative interview studies and PPI work, key stakeholders have expressed a desire for self-management tools. Generally there was excellent feedback to taking part in the study and being able to test preliminary versions of any self-management intervention that may support children with Perthes' Disease and their families. The focus groups pose different time and logistical burdens to participants. To mitigate these as best as possible, focus groups were carried out local to each site to prevent the need for excessive travel. The focus group was organised at their treating clinical site (albeit in a neutral space within the clinical site) and all travel costs were reimbursed. Similarly there was provision of childcare costs for focus group participants should it have been needed; this would have allowed for the care of additional children to have been arranged. This was not needed in any of the three focus groups. Focus groups also took place in the school holidays at the request of a number of potential participants which hopefully reduced burden for children/families taking part.

The research team did not identify any potential for serious adverse events (SAE) arising for participation in this study. However, to ensure the safety of the participants, a plan was established and discussed with each of the PIs for the study. There were clear instructions within the NON-STOP app to contact their treating clinical team if there were any cause for concern. If any safeguarding issues arose within the focus groups, the researcher was to report this to the recruiting clinician at the site and local NHS site policy would be followed. This procedure was well established with the PIs in the setup of the study, and thankfully never exercised as no issues arose.

6.10 Results

Between 29th January and 25th March 2024, 31 participants were recruited to the before and after observational study. A detailed breakdown of participant involvement in each of study components are provided in Table 6.4.

6.10.1 Before and after observational study

The total number of participants invited through existing orthopaedic appointments is shown in Table 6.4. It also outlines the staged approach to invitation, consent and onboarding of the NON-STOP app. While the sample size within this usability and acceptability study is not that of a definitive clinical trial, 86% of those invited to take part in the study consented, 72% of those invited engaged with the app. Follow up responses were low, and is explored in more detail in the discussion.

Table 6.4 – Participant engagement with the study

	n (%) from available population	**n (%) from initial sample**
Invited to take part in study	36	-
Consented to study	31/36 (86%)	31/36 (86%)
Completed NON-STOP onboarding	28/31 (90%)	28/36 (78%)
Engaged with app	26/28 (93%)	26/36 (72%)
Completed post-trial survey	20/28(71%)	20/36(55%)

The demographic representation within the study was fairly typical of the patient population (Table 6.5). Of the 36 participants who were invited, two were of Asian background and one agreed to take part in the trial. There was a pre-specified target of 20% female participants to align with epidemiological data [263], and 7/31 (23%) of the recruited before/after observational study participants were female.

Table 6.5 – Characteristics of participants in the before and after observational study

Characteristic	**Participants**	**National data**
Total number of participants consented	31	-
Age at diagnosis: mean years (range)	5.2 (2 to 10)	5.4 (median)
Age at time of recruitment: mean years (range)	7.8 (4 to 14)	-
Sex: n= male (%)	24/31 (77%)	77.6%
Ethnicity n= White British (%)	30/31 (97%)	90.6%
Ethnicity n= Asian/Asian British (%)	1/31 (4%)	-

**National data taken from epidemiological study by Perry et al [263].*

The NON-STOP app was used for six-weeks. In Table 6.6 app use is demonstrated using various metrics based on the process measures for the participants (n=26) that completed onboarding and engaged with the app.

Table 6.6 – NON-STOP app use during before and after observational study for participants (n=26) who engaged with the app

Measure	**Cohort total**	**Median per participant**	**Range**
Number of app log ins	254	5	1 to 37
App log ins week 1	67	2	1 to 7
App log ins week 2	39	3	0 to 7
App log ins week 3	43	3	0 to 5
App log ins week 4	31	1.5	0 to 6
App log ins week 5	31	2	0 to 6
App log ins week 6	43	3	0 to 6
Met target for progress reward week 1	12	N/A	N/A
Met target for progress reward week 2	8	N/A	N/A
Met target for progress reward week 3	6	N/A	N/A
Met target for progress reward week 4	5	N/A	N/A
Met target for progress reward week 5	3	N/A	N/A
Met target for progress reward week 6	4	N/A	N/A
Average (mean) pain score after six-weeks	2.05	1.85	0 to 10

The number of total app log ins reduced across the six week period. There was variability in levels of engagement across the sample, with some that did not use the NON-STOP app in certain weeks, and some that accessed most days.

The progress reward was linked to customisation of the avatar (Bobby the bone) in weeks one and two; this required participants to complete three activity sessions. This increased to four sessions in weeks three and four, and to five sessions in weeks five and six. As the trial progressed the number of total app log ins reduced, as did the number of times the progress target was met.

Collecting pain scores can be useful for interventions that may aim to improve, or at least consistently measure pain. The way that the pain data was used is slightly different in that it dictated the duration of exercises within the NON-STOP app, as described in Chapter five. The range of the pain score shown in Table 6.6 demonstrates that there were participants who experienced 'high' and 'low' levels of pain. This meant that they used each possible variation of the exercise length, i.e., some lasted 20 seconds, some lasted the full 60 seconds. The average pain score (both mean and median) can be used to demonstrate the most commonly recommended exercise length.

In Table 6.7, the exercises within the activity section are ranked in order of how many times they were used and the average times each exercise was used per week is presented. The scissor and Frog jumping exercises were only available in weeks five and six, and this is reflected in the activity usage data.

When using the app, participants needed to have logged in and used the app once a week for four weeks to unlock the 'Frog jump' and 'Scissor jumping' exercise. It was expected that they would be the least used activity due to being available for a maximum of two weeks. While it is difficult to determine the popularity of these activities, when the use is averaged over the weeks available, Scissor jumping appeared a relatively popular activity.

Table 6.7 – Activity use during NON-STOP app testing period

Activity	**Total times completed**	**Average times used per week**
Abduction	152	25.3
Crab Walk	142	23.7
The Butterfly	142	23.7
External Rotation	138	23.0
Extension	137	22.8
Flexion	126	21.0
Ninja Squat	111	18.5
Monster Walking	106	17.7
Scissor Jumping*	47	23.5
Frog Jumping*	36	18.0

**Exercises unlocked for weeks five and six of the study*

The available data on the use of the Learning section of the app was not as detailed in terms of available outgoing data, i.e. the exact number of times each section was accessed. App-use demonstrated that the most accessed sections were “What is Perthes’?” and “Nutrition”. “Lifestyle” and “Wellbeing” were the least commonly accessed elements of the Learning section.

Usability assessment

The scores from the Health ITUES are displayed in Table 6.8 and demonstrate the usability score for the NON-STOP app. To remind the reader, the score is out of five, with a higher score demonstrating a higher level of agreement with the statement. A cut off score of 4.32 has been previously validated as a cut-off score for ‘usable’ [276]. The scores for each element are provided for a more detailed interpretation of experience. However in the instructions for using the measure, the mean of each domain, and then the total score is to be used.

Table 6.8 – Health ITUES scores for the NON-STOP app

	Domain	**Mean score (out of 5)**
	Impact	***4.77***
1	I think the NON-STOP app would benefit persons living with Perthes' Disease.	4.9
2	I think the NON-STOP app would improve the quality of life of persons living with Perthes' Disease.	4.8
3	The NON-STOP app is an important part of meeting my information needs related to my health.	4.6
	Perceived usefulness	***4.64***
4	Using The NON-STOP app will make it easier for me to monitor and learn about my health.	4.8
5	Using The NON-STOP app will enable me to monitor and learn about my health.	4.75
6	Using the NON-STOP app makes it more likely that I will track my health.	4.6
7	Using the NON-STOP app will be useful for receiving reminders about my health.	4.75
8	I think the NON-STOP app presents a more equitable process for managing my health.	4.6
9	I am satisfied with the NON-STOP app for helping me monitor and learn more about my health.	4.65
10	I will monitor my health in a timely manner because of the NON-STOP app.	4.45
11	Using the NON-STOP app will increase my ability to track my health.	4.55
12	I will be able to track my health whenever I use the NON-STOP app.	4.65
	Perceived ease of use	***4.8***
13	I am comfortable with my ability to use the NON-STOP app.	4.75
14	Learning to operate the NON-STOP app is easy for me.	4.85
15	It will be easy for me to become skillful at using the NON-STOP app.	4.7
16	I find the NON-STOP app easy to use.	4.9
17	I can always remember how to log on to and use The NON-STOP app.	4.8

	User control	***4.25***
18	The NON-STOP app gives error messages that clearly tell me how to fix problems.	3.85
19	Whenever I make a mistake using the NON-STOP app, I recover easily and quickly.	4.25
20	The information (such as on-line help, on-screen messages and other documentation) provided with the NON-STOP app is clear.	4.65
	Total mean score for Health ITUES for NON-STOP app	4.63

All but two of the individual elements were rated over the cut-off of 4.32. The two statements that did not pass the threshold for 'usable' were related to user-errors and after users had made a mistake. Both of these elements score a majority of 'neither agree or disagree' (which scores '3') because error messages and mistakes were not a part of using the NON-STOP app. Overall, the usability score of the NON-STOP app was greater than the cut-off score, so could be termed 'usable'. The highest score achieved was in the 'ease of use' domain which produced a mean score of 4.8.

6.10.2 Integrating before and after observational study findings within the nested focus groups methods

The approach used here was an 'explanatory sequential' mixed method design [147]. The before and after observational study results were underwent preliminary analysis and the used to inform the content of the topic guide for the nested focus group (Appendix V). Questions in the topic guide were included to ask what participants' favourite parts of the app were and why. Similarly, questions exploring the optimal frequency of use were included i.e. in the future, how many times a week would be realistic to use the NON-STOP app for children with Perthes' Disease and their families? The coding matrix was modified in light of the trial findings, and used subsequently to support analysis.

6.10.3 Nested focus group

The nested focus groups took place at the three sites (Leeds, Alder Hey and Sunderland) that recruited participants for the before and after observational study. Of the 31 participants that consented to the before and after observational study, 25 agreed to receive the study information for the nested focus group and eleven consented to take part. Nine participants took part in the focus groups, two participants at Leeds, four at Alder Hey and three at Sunderland.

The focus group participant characteristics are described in Table 6.8. Following this, an overview of the themes and sub-themes are presented following the coding framework shown in Table 6.3. Illustrative quotes from participants are used to give context to the codes identified within the focus group data. A quote table which shows the full dataset is included as Appendix Y. A thematic table, which displays the frequency of each theme mentioned by participants is also included as Appendix Z.

Table 6.9 – Characteristics of participants in the nested focus group

Characteristic	**Value**
Total number of participants	9 (4 dyads, one parent)
Age at diagnosis: mean years (range)	4.4 (2 to 9 years old)
Age at time of recruitment: mean years (range)	6.9 (4 to 11 years old)
Sex: n= male (%)	5* (55.5%)
Ethnicity n= White British (%)	8 (100%)

**3 male children, 2 fathers. Remaining participants were 1 female child and 3 mothers.*

6.10.3.1 Usability

To understand the usability of the NON-STOP app the ease of use was explored, and users asked to identify any issues experienced while using the app. Participants recounted their experiences of using the app together with their child, and times when a level of independence from the child was observed.

Problems with use

The first sub-theme related to issues that participants faced when using the app. In focus groups conducted at separate recruiting sites, participants reported that when logged in to the app on two separate devices, for example on each of the parents' smart devices, rewards did not synchronise. One participant explained:

> "*if I was working or he was, you couldn't use it on different devices. Dad had it on his phone but then when he went on, he had to restart. If child already had, say, three stars that week, it didn't connect.*"
>
> *Mother of 4-year-old male*

A participant in a different group reported:

> "*It doesn't match up (sessions on different phones with same log in). Mum has more data on her phone than mine*"
>
> *Father of 7-year-old male*

This is certainly an element that could affect app use given that the intervention has been developed based on psychological theory that increases motivation through rewards for the children. Particularly important to consider the differences in family dynamics that can affect this with differences in home set ups and working patterns of the modern family.

A similar issue arose for one participant who explained that there were problems, at times, with reminders:

> "*Sometimes they (the app reminders) came through, sometimes they didn't*"
>
> *Mother of 5-year-old male*

Once again, considering the influence of psychological theory on behaviour change reminders/prompts were built in to the NON-STOP app to support participants when using it. As a result, it is reasonable to suggest that without them, use may be hindered.

Elements increasing use

There were certain elements within the NON-STOP app that participants described when asked what they liked and why. There were some elements that appeared to increase the use of the app, particularly from the perspective of how easy the child/family found the app to use. Instructional videos were useful to a number of participants. One child said:

> "*I liked copying the exercises"*
>
> *7-year-old male*

In another focus group, a parent also recounted how the app assisted with instructions to help the child use it. In the same conversation they also highlighted the impact of the NON-STOP app being entertaining. The in-built timer appeared to provide extra motivation to complete the exercises. They said:

> "*the app was showing you how to do it. Again, I felt like it was more of a game for him, it was more entertaining, it was like a race, how many can you do before the time runs out"*
>
> *Mother of 5-year-old male*

Demonstration of independence

The final sub-theme had particular relevance to the aims of the study. When considering how usable and acceptable a self-management intervention is, one must consider whether it can be used independently. There were a number of examples of participants recounting times when the child had used the NON-STOP app independently. One example is shown in an interaction between participants in a focus group:

> "*Father of male: And you found it really straightforward, didn't you? He would go on, pain today, and then he has a little scroll to see, and off he goes.*
>
> *Male, 7: yeah*

Mother of female: Yeah, that's the same as you isn't it

Female, 7: Yeah"

Interaction between 7-year-old male and father, and 7-year-old female and mother

Whilst it is a positive achievement for the NON-STOP app to be used independently by the children, independence was also important for family members. Over the course of this doctoral programme of work, there has been evidence of the impact that varied care has had on key stakeholders [155]. There has also been a strong need for reliable information as part of self-management. This was built in to the NON-STOP app in the Learning section. The impact of this was highlighted in a focus group:

"*I feel like it really allowed me and (father of child) to really get an understanding of Perthes' Disease"*

Mother of 4-year-old male

6.10.3.2 Rewards

Rewards made up a large part of motivating children to use the NON-STOP app. This was a conscious element of design and development, as described in Chapter five. It was apparent in the responses during the focus group that this had a meaningful impact on the participants when using the app.

Impact of rewards on app use

Customising Bobby the bone, the avatar, with accessories was discussed a number of times across the three focus groups. One interaction between a child and his father highlights that it was a key factor in their use of the NON-STOP app. They said:

"*Father: Why do you enjoy the app more? Because of Bobby, wasn't it? I think it was; the reward was getting to…*

Child: I like to customise Bobby the bone"

7-year-old male and father

Referring back to the psychological theory that underpins this thesis, autonomy and competence are vital to sustained changes in behaviour. It is possible to suggest that autonomy and competence in the context of the NON-STOP app led to a user getting their reward. If they could not use the app well, then they were not able to achieve this. One mother described the impact that the rewards had on the user:

> "*Even just getting the star, he would be happy, he is at that age where he's getting stars. He's like, "I got another star, I got this, I got that". He was going back telling my parents, he was proud of that accomplishment"*
>
> *Mother of a 5-year-old*

The impact of receiving stars for completing exercises on any given day did not only motivate the children. One mother described how the motivation was felt across the family:

> "*I think when we knew there were the stars, it's a goal for everybody and I think you need that in life, don't you, with everything"*
>
> *Mother of 4-year-old male*

It was common across the focus groups to hear parents discuss that the rewards had made it easier to maintain some consistency using the app.

6.10.3.3 Experience of the NON-STOP

To ascertain how acceptable and usable the NON-STOP app was, participants were asked to describe their positive, and less positive experiences of the app. Both are vital to further development of the intervention. It was also important to understand whether there was any progression during the app-testing period from users such as increased independence when using the app or doing other activities.

Positive elements of the app

Participants across the three focus groups described parts of the NON-STOP app that they liked. Highlighting things such as the colour schemes within the app being bright and welcoming. Also the interactivity of the app was mentioned a number of times. On a few occasions, there was a particular focus on how information had been adapted and tailored to meet the needs of the users. One parent explained her feelings towards the learning section where the Perthes' Disease stages are described, and the change of shape that the femoral head goes through is described using an ice-cream analogy:

> "*I thought it was absolutely brilliant, the way it was worded. There was more understanding because of the stuff that's online that you're trying to read up on is not very clear. And the way it described it as an ice cream, the way it melts and then. I thought, no-one's ever told me that"*
>
> *Mother of a 5-year-old male*

Children within one of the focus groups interacted, discussing why the NON-STOP app was acceptable to them. They said:

> "*Female, 7: I think it's (the app) just good for everyone.*
>
> *Male, 7: Yeah. Really fun"*
>
> *Interaction between 7-year-old male and female*

Less positive elements of the app

Two issues were raised relating to less positive elements of the NON-STOP app. They related to how many times users were required to use the app a week in order to get their reward. One mother said:

> "*Three times a week, fine. I think when it got to the last week, I think they had to do it nearly every day"*
>
> *Mother of 4-year-old male*

This was the case, users needed to complete their exercises five times a week in weeks five and six of the study in order to get their reward. A similar theme is discussed towards the end of this section.

The other element of the NON-STOP app that was mentioned in a less positive way was the reminders. They were inconsistent amongst users. Some had reminders every day, some had none at all, and some were somewhere in between. A participant reported:

> "*Sometimes they came through, sometimes they didn't"*
>
> *Mother of 5-year-old male*

As discussed earlier, prompts/reminders were intentionally included as part of the intervention development. It was not made clear during the focus groups why notifications may have sometimes been delivered, and other times not. Users were prompted on their first use of the app to enable these notifications, and could disable the notifications, but variability within the user's experience is an area for consideration. This would need refining in any further development of the NON-STOP app.

<u>Progression throughout the app-testing period</u>

The final sub-theme explored relates to the progression that users demonstrated throughout the app-testing period. This relates not only to using the app, but also the progress that users made outside of app-use as their Perthes' Disease progressed. One mother explained the increase in competence over the six-week period:

> "*I think the videos really helped; we watched the videos first. I think obviously as the weeks went on and it increased, we didn't need to watch the videos as much"*
>
> *Mother of 4-year-old male*

As children progress, it is reasonable to expect that they may not need the same level of interaction with the NON-STOP app. It was clear that there were still uses for

elements of the app as children progress. This is demonstrated by a father explaining how the Activity section of the app was still a part of their routine despite the child's progress. He said:

> "*because he's missed doing things like football, and being able to go on things, he hasn't been able to, and he's kind of, well into that now, so it's kind of, a bit of a transition thing for him. But it's still keeping him doing his stretches, and his good movement"*
>
> *Father of 7-year-old male*

This is of particular importance when considering the implementation of an intervention like the NON-STOP app and considering who it is applicable to.

6.10.3.4 Comparison to previous and existing care

The aim of developing the NON-STOP app was never to replace elements of care such as physiotherapy or advice from professionals. It was, rather to supplement existing care and provide the tools necessary for children with Perthes' Disease and their families to self-manage. To consider the intervention in the context of the participants' lives is in line with the aims of this project. To do that, participants discussed the NON-STOP app in comparison to the care they have received in the past, or were receiving at that moment. Some participants explained the impact of using the app in relation to the stage of Perthes' Disease they were at when the focus group took place.

Relationship to current care

The first sub-theme related to how the NON-STOP app fits in to participants' lives and what impact, if any, it had on things like physiotherapy care or completion of home exercise programmes.

One mother explained the benefits that using the NON-STOP app had on her attending the hospital for appointments. She explained:

"*He was actually seeing (local physiotherapist) every two or three weeks, and now it's every four weeks"*

Mother of 5-year-old male

As discussed, aim of the NON-STOP app was to supplement existing care and give users the tools necessary to self-manage. Giving advice and education on managing a condition is a key part of many healthcare professionals' care. The NON-STOP app has led to a reduction in appointments needed for at least one participant.

Supplementing care is not only done by recreating the regime using a different medium. In one focus group, the mother of a child explained that the exercises in the NON-STOP app were slightly different to those prescribed by the local therapist. They were then incorporated into the child's regime and provided some alternative choices to maintain engagement. The mother highlighted how the input was integrated into their regime:

"*We do that including our physio, so we do both. The app is slightly different from the exercises she gets from physio"*

Mother of 7-year-old female

<u>Differences to previous care</u>

Honest reflections of previous care were given, and discussions within the focus group described common themes around how the app was "more fun" and provided a place for information that was in keeping with the modern day. One mother explained how it was better than previous methods for advice on exercises:

"*I definitely think he got more benefit from the app than us looking at a piece of paper and trying to figure it out, where the app was showing you how to do it."*

Mother of 5-year-old male

During one discussion, a father explained that local resources and access to care were impacted. The variation of care that exists in the professional world had impacted their care and the relationship with their local therapist. He went on to explain how access to the NON-STOP app may have been beneficial earlier:

> "*he lost his hydrotherapy. We didn't get on with the physiotherapist, they said there was conflicting views, and things, and she was trying to influence the school, and things, and it went a bit crazy. So, (child) has had some physio at another centre, but if we had the app earlier, it would have been really, really, really useful.*"
>
> *Father of 7-year-old male*

Relevance to stage of Perthes' Disease

Children at different stages of Perthes' Disease require different levels of input and their care has different focuses. For example, from Chapter four, a recommendation stated that in the early stages of Perthes' Disease, high-impact activities should be avoided. In contrast, in the later stages, i.e. when remodelling, activities should only be limited based on pain levels. In the focus groups, participants explained that the NON-STOP app was useful in both stages. Similarly, children earlier in the stages of Perthes' Disease are likely to be younger, where factors such as understanding and compliance can affect users' experiences.

A mother explained that her son had found some exercises easier than others, and that perhaps his age had influenced this. She said:

> "*The frog jump, yeah. I think a couple (he found hard), he got better, but I think some of them, because of his age, some of them he probably didn't do as well as obviously somebody that's a bit older.*"
>
> *Mother of 4-yar-old male*

A father in a different focus group explained how his son had found that the NON-STOP app was no longer the main focus of his care. Rather that activity had become his key focus. Nevertheless, there was still a role for self-management and engagement with elements of the NON-STOP app. He explained the relationship between his son and the app in the later stages of Perthes' Disease:

> "*now, because he's missed doing things like football, and being able to go on things, he hasn't been able to, and he's kind of, well into that now, so it's kind of, a bit of a transition thing for him. But it's still keeping him doing his stretches, and his good movement"*
>
> *Father of 6-year-old male*

6.10.3.5 The future

Participants discussed the functionality of the NON-STOP app and changes that may be needed to increase the acceptability and usability of the intervention. As well as this, ideas for improvement and continued engagement were given.

Changes to functionality of the app

The first sub-theme relates to what changes may be needed when optimise the app usability. As mentioned in the first theme, having an account that syncs regardless of which device it is used on is important. This was highlighted by one participant:

> "*I think if you've got two parents that are doing it together or you've got, say, an iPad and a phone, then I think it's probably good if that could work"*
>
> *Mother of 4-year-old*

Another discussion point related to the Learning section; this section was made up of text and links to external resources. It was suggested that changing these to videos with captions and animations may make the sections more user-friendly. In a conversation within the focus group, two parents discussed:

> "*Father: Yeah, I think having videos in the learning section would be good, yeah*

Mother: Yeah, it could be useful"

Interaction between father of 6-year-old male and mother of 5-year-old male

When asked about this in another focus group, a participant said:

"*I would say, definitely for him, because he's reading but he wouldn't be able to read a full paragraph of what it's all explaining"*

Mother of 5-year-old male

This mother was considering the accessibility of the child in relation to the app. It is possible that by making information accessible to children with text alternatives for those who want it, there could be increased engagement.

Ideas for the app

A common trend in the responses when asked about what could be done to maximise motivation and engagement was around making sure there are new customisation options for the avatar, Bobby the bone. Ideas from child participants in the focus groups gave ideas such as "Bobby the pirate", "Bobby as a rockstar", "Bobby as a footballer" and seasonal outfits like "Santa and Easter Bobby". Integrating the ideas of the target population is vital when aiming to maximise usability and acceptability of this novel intervention.

Other ideas related to progression as the child gets older. One participant suggested more exercises as the child gets older to keep them from getting bored. Another described that a notes section would have been useful. Initially that was thought to be for sharing information with professionals, but actually the parent explained this would be for personal reflection primarily. She said:

> "*I would've liked to have something on where I could write in when he's had a bad day, so I could document it all in one area. And looking back on where it brings the faces on how he's feeling on that day, to go back and see what that is"*
>
> *Mother of 5-year-old male*

A discussion point was raised about increasing engagement in the wider patient population. When discussing long-term use of the app and how engagement could be considered, a mother explained that the clinical setting could be used as a perfect place to share information about the intervention. They explained their experience of finding out more in their clinic:

> "*you've got the QR code in the clinic, and I think that's got the guide on, hasn't it, of what the app is. That might be something you could share. Because that's where people are sat. You're sat, you're waiting, you've got your phone, obviously you're there for that reason on that day for your child with Perthes' and you're like, look, there's that. That, especially if parents haven't heard of it"*
>
> *Mother of 4-year-old male*

6.10.3.6 Additional points for app use

Two additional points were raised in the focus groups relating to the app use. The first related to how the app had been used to educate others about the condition and about what self-management involves. The second related to what the optimal dose for the intervention, i.e. how many times a user needed to engage to receive their progress reward.

Educating others

The NON-STOP app was designed for children with Perthes' Disease and their families. In the study, many participants recounted their experience of using the app

in other environments and domains. Users recalled showing external parties such as schools and sports clubs the NON-STOP app and using it to explain their condition. One mother explained how she had used it to educate friends and family:

> "*I really liked that, because obviously, we get a lot of, well what is Perthes, and I would just be like, look, have a read, you know what I mean? Yeah, show the, like, school teachers, and like, obviously, family, and stuff like that, who would, yeah, like my sisters, and stuff like that, I'd be like, they'd be like, so what actually is it, and you'd be like, there you go, have a look at it, have a read.*"
>
> *Mother of 7-year-old female*

Two parents at separate focus groups explained the benefit of having the child's school look at the Learning section of the app. One explained how it resulted in engagement with classmates:

> "*The school thought it was absolutely brilliant, the way it was explained. I was going to try and get them on the app as well, to try, because they will do his exercises with him as well*"
>
> *Mother of 5-year-old male*

A father explained that the app was not available when they were diagnosed, so had to use other methods to explain the condition to school and this may have led to negative experiences for his son. He described the impact it could have had:

> "*it would have been useful for us to have that to show his school, and maybe they could have done, used the app themselves in school, when he was missing out on things in PE that he couldn't do, and whatnot*"
>
> *Father of 7-year-old male*

Intervention dosage

At various points in the data collection, the frequency of exercise completion was discussed. Intervention dosage as a concept of intervention fidelity is explored in more detail in the discussion section. There are however practical implications to also consider when suggesting the use of an intervention. In the focus groups, participants made their feelings known regarding acceptable frequencies. One mother said:

> "*Our aim is we do it twice a week because I do feel that it is really helping.*"
>
> *Mother of 4-year-old male*

This experience of improvement or feeling of self-management is important, particularly when combined with the experience that was shared earlier which suggested a sense of 'pressure' to complete it almost every day.

In a different discussion, a parent reported that the number of times the NON-STOP app was used could be of benefit to try and ascertain whether the child was in a period of discomfort. Many parents explained that their child may not always be forthcoming with that information due to feat of activity restriction. A mother explained:

> "*It would be a good way of looking back again and seeing how when he was been able to complete it. Because there were times when he felt like he couldn't use it, like, Mum, I need to rest"*

The number of times recommended was a measure of usability and acceptability in this study. It is a point for consideration and discussion when planning wider implementation of the NON-STOP app.

6.10.4 Synthesising the mixed-methods results

The mixed-methods approach within this usability and acceptability study provided an evaluation of the NON-STOP app. Section 6.10.2 described how the before and after observational study data was used to inform focus group topic guides. Qualitative data was then used to gain more insight into what parts of the NON-STOP

app were fun and engaging. It was also possible to ascertain why those parts worked on a more detailed level. For example, a strong theme throughout the nested focus group was the impact that rewards had on the use of the app, and in particularly the high dose required to progress to weekly rewards acted as a disincentive. It would not be possible to evaluate impact that the reward process had on use using the quantitative data alone.

Another insight from the mixed-methods synthesis was around app engagement as time went on. In the focus group more detail on intervention dosage and how often the app was used was given. This was not an aim or objective of the study, and came organically from the data within the qualitative study. It was clear from the quantitative data that engagement reduced with time. Collecting this data in the qualitative arm of the study allowed the researcher to explore why this happened. Demonstrating how the mixed-methods approach allowed for a more nuanced understanding of the positive elements of the NON-STOP app, and areas for development.

6.11 Discussion

In this section, a summary of the main findings is presented from both the before and after observational study and the nested focus groups with an outline of how the mixed-methods approach influenced the findings. After this, the strengths and limitations are presented relating to the study. Before moving on to the conclusion, the positional reflexivity of the researcher is explored, as has been done in previous chapters.

6.11.1 Main findings

Mixed-methods were used to test the usability and acceptability of a novel, digital self-management intervention for children with Perthes' Disease and their families. The objectives of this study were achieved, and quantitative and qualitative information were analysed relating to the NON-STOP app. Study procedures, such as

recruitment processes and follow up were, also explored prior to a definitive clinical trial.

In the before and after observational study, process measures demonstrated a good level of engagement from those who completed the onboarding (training package). With 93% of participants going on to engage with the NON-STOP app. Over the course of the six-week period, engagement declined, with app log ins reducing towards the end of the testing period. Similarly, the number of participants who met the requirement for the rewards also reduced. Making sure that rewards can be realistically achieved by users of the NON-STOP app is important for long-term use. Particularly given that the inclusion of rewards in the NON-STOP app has been based on one of the psychological theories that underpins the project (behaviour change) [145].

It is possible that the number of times the reward target was met reduced in the study due to the frequency needed to achieve the reward being unachievable for participants. Intervention dosage was an emerging theme from focus groups; participants discussed how by the end of the testing period, needing to use the NON-STOP almost every day to get the reward felt unachievable.

Study procedures such as outcome collection and recruitment and retention rates were explored in this feasibility study. The before and after observational study gave an indication of collecting outcome data at two time-points. From a methodological sense, this study demonstrated an ability to recruit participants for the before and after observational study that are broadly representative of the patient population (as seen in Table 6.5). There is a risk of selection bias for engagement with the app as a result of the recruiting clinicians being asked to recruit purposively rather than consecutively.

The before and after observational study was also able to gain an insight in to how users found the NON-STOP app in a practical sense. Whilst the sample size was designed to produce statistically significant results, the usability score (Health ITUES) did provide early indication of a usable digital intervention for children with Perthes' Disease and their families. All but 2/20 elements met the cut-off score for being found

to be ‘usable’ by the participants. The two that did not meet the cut-off score were not applicable to this project (related to mistakes and error-messaging).

The phenomenon of intervention dosage is considered in the literature, particularly in work from Borelli [285]. The concept of intervention dosage refers to the amount and frequency of an intervention that participants are exposed to. In the context of the NON-STOP app, this would include number of sessions, duration of each session, and the overall length of the intervention period. Borelli emphasizes that an optimal dosage is crucial for achieving the desired outcomes without overburdening participants which can result in disengagement. In this usability and acceptability study, it is possible to suggest that five sessions per week led to overburdening of participants. This could explain the reduced engagement as demand increased. It is also possible that as children/families became more familiar with the intervention, they got to know their exercise regimes and completed them without the use of the videos on the NON-STOP app. Intervention dosage for the NON-STOP app, i.e. the number of engagements per week that are needed to receive the reward, would need careful consideration prior to future testing. For instance, in a definitive clinical trial, in order to maximise the effectiveness of the intervention, whilst also maintaining participant engagement.

In the qualitative study with key stakeholders in Chapter three and also in the clinical consensus recommendations study in Chapter four, a strong need for education, advice and reliable information was identified. This was built in to the NON-STOP app in the Learning section. From the before and after observational study, participants accessed certain parts of that section more than others, with the majority of users engaging with the “What is Perthes’?” section. How useful this was as a resource, where it was useful, and why it was useful was highlighted in the nested focus group. Participants explained that the information was well presented, they enjoyed the colourful layout with pictures, and said that the information was delivered in an acceptable way. Many described how it had been used not just within its intended recipient group, the Learning section was shared with schools and sports clubs as well as wider family groups. Sharing the information to inform others of the condition appeared to be a key use for the Learning section.

Educating people involved in the lives of children with health-conditions has been studied previously. In a systematic review in 2022, Lancaster et al summarised the findings from studies that evaluated peer support programmes that aimed to improve quality of life and wellbeing scores for children and families with long term health conditions [286]. The review highlighted that school-based interventions which inform school staff and peers about the child's condition can lead to a more supportive and understanding environment. The NON-STOP app has the potential to be used in a similar way, as discussed by participants in the nested focus group. The app was used to educate wider-family members, teachers and other stakeholders about this rare condition. Some children even reported that peers had joined in with exercises at school. An improvement in integration and acceptance for the child could possibly lead to sustained engagement with the NON-STOP app.

In focus groups participants expressed how instructional videos and rewards were positive influences on app use. Children highlighted how the use of Bobby the Bone as a character allowed them to feel represented in the NON-STOP app. Being able to identify with the intervention aligns well with the theoretical underpinning, particularly SDT in which 'relatedness' is a key component of optimising motivation. The mixed-methods approach used in the study allowed the researcher to understand this in more detail. The approach also allowed the researcher to get a better understanding of not only how many times the app was used, but by whom. In the focus groups, many parents, and many children recounted their experiences of using the NON-STOP independently. Having children independently use the NON-STOP app aligns well with the SDT which looks at three main aspects of motivation that can lead to sustained behaviour change [105]. These are discussed in relation to the NON-STOP app in Chapter five, however, to summarise based on the findings, independent use of the NON-STOP app demonstrates a child's autonomy and competence to use the intervention.

6.11.2 Strengths and limitations

The mixed-methods approach is a significant strength of this study. It allowed for a comprehensive evaluation of the usability and acceptability of the NON-STOP app. It did so by integrating the quantitative data from the before and after observational study, using that to formulate the discussion points of the nested focus group. The before and after observational study provided objective metrics on app usage, engagement levels and specific outcomes such as pain scores and activity completion rates. The nested focus group complimented this data by giving a meaningful understanding of why these metrics may have occurred. Participants used the qualitative arm of the study to share their personal experiences of using the NON-STOP app. They highlighted positive elements of the app as well as elements that were challenging and which might lead to refinements in the app content. These challenges in particular, are essential when considering wider implementation or testing of the intervention.

Possibly more important than the identification of challenges, participants also provided useful insight as to how they might address these challenges and improve the app. To give a practical example of how the mixed-methods approach addressed the aims and objectives of the study effectively. Participants reported that the daily reminders built in to the app sometimes came through and sometimes did not. The before and after observational study process measures could not capture this. However by using mixed-methods, there is meaningful data that helped the researcher firstly know that it happened, and secondly understand how to address it in the future. This could be done by reporting back to the app developers, who can explore reasons why the reminders may not have worked. It is then possible to look at how to improve this for the next iteration of the intervention and maximise engagement. Whilst it is not possible to draw large assumptions from the data given the small sample size both in the before and after observational study and the nested qualitative focus groups. It is reasonable to consider that the experiences of using the NON-STOP app are likely to be similar across the cohort. Particularly when thinking of things like functionality of the app and reminders being sent as desired.

A key strength to the project as a whole is the collaboration with the developers of the digital self-management intervention. The researcher has taken the lead role in each step of the intervention design and development in relation to content, layout and testing. With a particular focus on this app-testing study, the level of data extraction demonstrates how collaboration and input from the app developers can lead to meaningful outcomes. The collaboration ensured that the aims and objectives of the study were met. The granular details that are possible to extract were unknown to the researcher prior to the collaboration. Thanks to the professional background and expertise of the developers, it was possible to assess things such as the popularity of each exercise on the NON-STOP app. As discussed earlier, combined with tools such as the TIDieR checklist, this can potentially demonstrate the elements of the NON-STOP app in detail in future research.

As discussed in the main findings, it is a strength of this feasibility study that the researcher was able to recruit participants that were generally representative of the patient population. Similarly, the rate of recruitment and agreement to take part in this study is positive when considering future research which should involve randomisation and consecutive recruitment. This will allow for analysis for certain characteristics which may influence engagement. For example, in this study, the mean age of the participants who tested the NON-STOP app was 6.9 years. That age is fairly typical of a child diagnosed with Perthes' Disease, and accounts for the vast majority of the children who may benefit from the self-management information displayed in the way it has been in this version of the intervention. In the future, further exploration of usability and acceptability of the NON-STOP in different aged users would be recommended. Whilst this does not account for a large proportion of patients with Perthes' Disease, it is important to ensure that any intervention meets the needs of the intended users.

Testing the usability and acceptability of the NON-STOP app had limitations. A key limitation in the before and after observational study relates to the collection of follow up data. The proportion of responses that were gained is not necessarily low at 71%. However it required multiple reminders to participants to complete the survey and was not inexpensive in terms of time. Careful consideration would be

needed on how to mitigate this in the future. One potential is the introduction of incentives to complete follow up data. This is utilised in many studies to good effect [287]. In a recent meta-analysis, Abdelazeem et al reported a statistically significant increase in the response rate from participants when they were offered incentives (monetary or otherwise e.g. gifts). Additionally, the three sites used (Leeds, Sunderland and Liverpool) were all in the North of England. Despite the spread from west to east, a potential limitation is that it may not capture a diverse population in terms of socioeconomic diversity, ethnic background or service provision.

Whilst the participants that took part gave honest reviews and provided rich, meaningful data regarding the usability and acceptability of the NON-STOP app, the number of participants was much lower than hoped in the focus groups.

The first focus group, in particular, was quite different to what was planned due to a number of factors. Initially there were four dyads that had agreed to take part in the focus group. One of whom contacted the researcher to explain the child was non-verbal and that a face-to-face focus group was concerning to them, and they would prefer not to be involved. Another parent contacted the researcher to say they were no longer able to make it due to a family bereavement. A third dyad was unable to make it on the day, with no contact made regarding non-attendance. This left one dyad who took part in the first focus group, meaning that data collection took place as an interview rather than a true focus group. There are obvious limitations associated with this based on the advantages of conducting focus groups rather than interviews. These relate to things like participants not being able to discuss experiences and encourage natural conversations amongst people who had used the intervention [280]. However there is also an argument to suggest that having the parent and child in an interview may have encouraged a more honest reflection of the experience. With the lack of others present providing less reason to not share some aspects of care or experience. This also represented a protocol deviation from the qualitative data collection method. This presented the researcher with a situation that required input from ethics and governance authorities. After undertaking the dyad interview, contact was made with the REC who advised seeking advice from the sponsor of the study (University of Leeds). This was done and it was deemed that

collecting the data using the interview format, using the same topic guide, was in the best interest of the participants and acceptable. It meant that the participants who had travelled to the site for the focus group had not wasted their time, and the data could still be used in the study.

Attendance in general across the three focus groups was less than anticipated. Attendance was also largely, not wholly, from those who engaged regularly with the app, and had a lot of positive comments about the use of the app. Focus groups were selected in an attempt to encourage discussion between participants, and generate discussions that would provide insight into things such as potential refinement. The limitation of this choice however is that it is possible to suggest that participants who did not engage with the NON-STOP app were unlikely to attend a face-to-face focus group given that they did not engage with the app during the testing phase. This could be because they did not want to address their lack of engagement, perhaps due to guilt. Future research could adapt the methods slightly and use methods such as telephone interviews with low-engagers to try and gain meaningful insight as to why they did not engage with an intervention.

The final study of this doctoral programme of work continued the alignment with the theoretical underpinning of the project. From the design of the study, through to data analysis, the study was well grounded in the psychological theories SDT and SEM. With specific consideration of the mixed-methods approach, the study was able to assess behavioural outcomes such as app usage and engagement. It was also possible to assess the contextual factors that influenced app use, providing a holistic view of the intervention's acceptability and usability.

6.11.3 Considering the reflexivity of the researcher

As was done in Chapter three, it is necessary to consider the researcher's positional reflexivity. To reintroduce this concept, positional reflexivity involves a critical reflection of the researcher's own perspective in terms of beliefs and values. The influence of these on the research must then be considered.

In Chapter three, there were discussions around how the researcher's experience as a children's physiotherapist may have influenced the interviewing. For example, active steps were taken to avoid clinical discussions and allowing participants the time and space to respond. In this mixed-methods study to test the usability and acceptability of the NON-STOP app, these aspects were considered. Feedback with members of the supervisory team (SR, HJS) were once again utilised to offer an opportunity to reflect, but also to build from group to group in the qualitative study.

Considering the researcher's position in the before and after observational study. There was a potential for overlap between clinical responsibility and the research role within the study. For example, a potential risk of influence on adherence with children/families known to the researcher/children and families were recruited from three sites, one of which was where the researcher continued to practice. To try and minimise influence, the researcher did not bring up the testing of the NON-STOP app in clinical appointments, but was sure to be receptive to children and/or family members that wanted to discuss. This is in the interest of maintaining a good relationship with the child and the family, as is the researcher's standard of care. As with the nested focus group, regular discussions and opportunities to feedback with the supervisory team were taken.

Continuing to be reflexive allowed the researcher to maintain a conscious awareness of how positionality influences the research. By doing so, the potential impact on the rigour of the research is mitigated. Another step to ensure the influence of the researcher's position was not overly influential was the involvement of the independent facilitator in each of the focus groups. This individual, whilst a children's physiotherapist, was not involved in the same clinical service as the researcher. Taking steps like these allowed the researcher to offer a nuanced interpretation of the findings.

6.12 Conclusion

The mixed-methods approach used in this app-testing study provided valuable insights into the usability and acceptability of the NON-STOP app. Through a before and after observational study, and a nested focus group, the findings demonstrate that the NON-STOP digital self-management intervention for children with Perthes' Disease and their families is both usable and acceptable. There are some caveats regarding long-term engagement, and these should be addressed by refining the app content prior to a definitive clinical trial. The methods allowed the researcher to explore behavioural outcomes from both quantitative and qualitative measures. Engagement over time was demonstrated using metrics drawn directly from the app. The metrics suggested that further refinement of the NON-STOP app's reward structure and dosage of use would be beneficial for maintaining acceptable levels of engagement over longer periods.

Through the nested focus groups, it was possible to gain a deeper understanding of participants' needs. Particularly around the educational content of the app, both in what is included and who it is tailored for. The feedback from both children and families has been instrumental in shaping the ongoing development of the NON-STOP app. Overall, the study contributes meaningfully to the optimisation of the intervention, and provides a strong foundation for its application in a larger clinical trial.

Chapter 7 – Discussion

7.1 Introduction

The final chapter of this thesis discusses the project as a whole. Firstly, an overview of each of the included elements of the project is presented. After this, the strengths and limitations of the doctoral programme are discussed. Key points for discussion are outlined, followed by the impact of the project in terms of practice and policy. Next the findings are summarised, and the aims and objectives are revisited. The thesis concludes with recommendations for future research based on the findings of this work.

7.2 Summary of the thesis

A mix of methods were used in this project to develop the NON-STOP app, a digital self-management intervention for the non-surgical treatment of Perthes' Disease. The methods employed were rigorous and evidence based. More than this, the methods were underpinned by two key psychological theories (SDT and SEM) that highlighted the importance of motivation leading to sustained changes in behaviour. The commonality between the two theories in context to this project was related to the importance of empowerment for children with Perthes' Disease. Maximising the autonomy and competence of children, increasing their ability to self-manage was at the forefront of this project.

Chapter three: Exploring the understanding of key stakeholders

Qualitative interviews with children with Perthes' Disease, their families and clinicians who regularly care for them were the first step in intervention planning. The study explored experiences of care, both positive and negative, and highlighted the hopes of key stakeholders in terms of future care. Participants highlighted the need for clinical consensus, and clear education amongst other key themes. Families, as

well as clinicians, were able to demonstrate the nuances of variation that exists in clinical care, that had previously been described by our research team [28]. Participants also offered insight as to what self-management should include. It was at this point that the idea of the NON-STOP app was first introduced to participants. An app had been identified as a potentially suitable mode of delivering self-management in PPI activities prior to undertaking this doctoral programme of work. Participants in the study offered their opinions and ideas as to what a digital intervention should include. These suggestions came from both clinical participants and child/family participants and focused on what they believed would motivate users to engage with a digital self-management intervention. Of particular relevance to the overall project, this study demonstrated the origin of rewards and gamification within an app in order to motivate children with Perthes' Disease to engage with the NON-STOP app.

The findings of this study were peer reviewed and shared with the clinical community in a published article in the Bone and Joint Open journal [155]. They have also been presented both nationally and internationally at relevant professional conferences. The results of the study were shared by email with all participants using infographics and summaries with links to resources such as the study website. The wider children's orthopaedic community was also informed of the findings using social media with links to said resources and journal articles.

Chapter four: Clinical consensus recommendations for the non-surgical treatment of Perthes' Disease

It was clear from the qualitative study that there was a lack of agreement amongst clinicians regarding the non-surgical treatment of Perthes' Disease. In the absence of robust evidence to direct practice, achieving clinical consensus was a reasonable next step. A two-round, modified Delphi study took place. In the first round, a survey designed with an advisory group that included experts in children's orthopaedics was shared with participants. The study involved two rounds of surveys completed by UK-based children's orthopaedic specialists including surgeons, physiotherapists and nurse-specialists with experience of treating children with Perthes' Disease.

Participants stated their level of agreement with the statements and had the opportunity to propose further statements for round two. Eighty-seven statements in total were included in the two rounds, and 45 achieved consensus and were presented as final clinical consensus recommendations. The consensus recommendations covered strengthening and stretching exercises, activity modification, pain management, mental wellbeing and referral to specialist services. The findings of this study offered clinical guidance in the absence of robust evidence, which in turn offers a potential to reduce variation of care. As well as reducing variation, this study underpinned the development of a digital self-management intervention. The content of the NON-STOP app was derived in part, by the recommendations produced in this clinical consensus study.

As in Chapter three, the findings of this study were written up and published in a relevant medical journal, the consensus study was published in the Bone and Joint Journal [219]. Here it was selected as a focus piece, which allowed the researcher to share key findings using video and other media. An infographic for this piece of work was also produced to promote the findings to the clinical community, including children and their families (Appendix M). This was designed with PPI collaboration and delivered a reading age appropriate for all. Copies were posted to clinical centres around the UK. The infographic is also hosted on the STEPS Worldwide charity website, so it is accessible for parents/families. The study findings have also been presented at national orthopaedic conferences.

Chapter five: Development of the NON-STOP app

Chapter five describes the design and development of the NON-STOP app. A digital self-management intervention for children with Perthes' Disease and their families. The app combines the findings of the first two studies to act as the vehicle for delivery of the clinical content, as well as the content derived from PPI activities. The design and development followed a combined approach of intervention development methods, all rooted in both theory and evidence. Namely, a combination of the Medical Research Council (MRC) framework for developing complex interventions

and the Behaviour Change Wheel (BCW). The combination of approaches ensured that the content of the NON-STOP app met the needs of the users, whilst still supporting behaviour change.

Features within the NON-STOP app included cartoon characters instructing children how to complete strengthening and stretching exercises. An avatar, Bobby the Bone, was customisable when children met their progress goals, which aligned with the reward element of the psychological theory which underpinned the intervention. The app also incorporated educational content for children and families to review. Collaboration with digital health app developers was vital to the success of this work, as was PPI and user-testing to ensure the content designed was accessible to the children using it.

The approach taken in developing the NON-STOP app is robust, the methods that were applied were rigorous and are accepted in the intervention development population. The methods used to design the NON-STOP app, which are paramount in the development of complex interventions prior to testing in a clinical trial, have been submitted for peer-reviewed publication.

Chapter six: Testing the usability and acceptability of the NON-STOP app

Mixed-methods were used to test the usability and acceptability of the NON-STOP app. The first component of the mixed-methods study was a before and after observational study which recruited from three UK sites. Thirty-one children with Perthes' Disease and their families were given access to the NON-STOP app for six-weeks. Data from the before and after observational study focused on use and engagement with the app. Looking at metrics such as number of log ins, pain scores, and which areas of the app were used most. Outcome measures that focused on usability of the intervention were completed at follow up.

The second component of the study was the nested focus group. In this qualitative arm of the study, participants from the before and after observational study were invited to take part in a focus group. The aim was to build on the findings from the

before and after observational study. Providing an insight into user experience and identify positive aspects of the app, as well as areas for improvement. The focus group provided deeper insights into usability challenges and also provided an opportunity to explore elements of self-management such as intervention dosage. For instance, through the before and after observational study it was possible to see how many times a week a child used the NON-STOP app. In the nested focus group, participants discussed why it was a certain number of times, and how many times a week was acceptable to them. Particularly when there was a reward element to the intervention.

Overall, the NON-STOP app was well received, and it was possible to understand the usability and acceptability of the intervention from this study. Engagement reduced over time; however, children and families gave valuable suggestions regarding refinement to maximise engagement.

Chapter six builds on the findings from Chapter four by providing the first phase of evaluation of an intervention based on methods that have aimed to reduce care that was varied [28] and deemed, by key stakeholders, as sub-optimal [155]. The study findings have been presented in a summary to participants via email and the early results of the study have been presented at national conferences, ahead of formal write-up and publication following peer review.

7.3 Review of exploratory hypothesis

In the first chapter of this thesis, an exploratory hypothesis and the aims and objectives were outlined. The hypothesis for this doctoral programme of work was that through a mix of methods, it would be possible to develop and preliminarily test a digital self-management intervention for the non-surgical treatment of Perthes' Disease. Using a range of methodologies and outcomes, summarised in section 7.2 above, and in each of the empirical chapters, this hypothesis has been explored and addressed. Refinements have been made to the intervention based on the findings

from the mixed-methods study, as well as PPI stakeholder input and relevant clinical trial experts.

7.4 Interpretation of main findings

The results from the studies within this doctoral programme of work offer novel conceptual insights. There is a lack of robust evidence for many children's orthopaedic conditions that warrant investigation. It is possible that this project, which enhanced the understanding of Perthes' Disease, produced clinical consensus, and developed an intervention to support care, could offer an approach that is transferrable to other areas. Particularly given that, to the researcher's knowledge, there are no methodological approaches that have been tested in children's orthopaedics, specifically in children's rehabilitation.

One key concept in this project is self-management. The theories employed throughout this thesis have been useful in supporting the development of a self-management intervention. SDT and SEM focus on increasing motivation to sustain a change in behaviour. [78, 109] In Chapter three, participants shared their desire for independence when managing Perthes' Disease, and explained that a digital intervention was something they would like to explore further. It would be possible to advocate using qualitative methods to outline what is important to key stakeholders in any condition in the future. Particularly given that the MRC Framework highlights the importance of involving key stakeholders in the design of complex interventions [44]. There are more nuances to self-management, and as a concept, it is not without its risks, which include disengagement through over stimulation, and lack of engagement leading to patients receiving no management as opposed to self-managing. These points are discussed in more detail in section 7.5 with specific focus on how the notion of self-management has been perceived in relation to the NON-STOP app, but also how it may be perceived in the wider children's orthopaedic setting.

The development of the NON-STOP was collaborative, specialists in app-design brought ideas from the qualitative study as well as the clinical consensus study. PAG members contributed to the design, with children providing practical examples of how to make the app fun and engaging. Valuable insight can be taken from what has been learned by the researcher in this project, and potentially be exported to other aspects of clinical research. Involving key stakeholders, PPI and PAG members offered a level of relatedness to the user, which, once again aligned well with the psychological theory, namely SDT, that underpinned the project. The identification of Bobby the Bone and the process of engaging with the app to earn rewards to customise the avatar was a positive element of the intervention. Children with Perthes' Disease and their families explained in focus groups that this motivated them to use the app. Identifying an element of an intervention, in the design and development stage was useful here, and is something to consider in any intervention development in the future.

There are some limitations, for instance, conducting a Delphi study in the absence of robust evidence is an acceptable method of achieving clinical consensus. On reflection, it was the most appropriate way of creating clinical content, as well as providing clinical guidance for a professional group where one had previously not existed. It is however important to understand that in the future, guidance may need revisiting should robust evidence arise. For instance from a definitive clinical trial. This would be vital in terms of updating clinical communities, but also ensuring that an intervention remains relevant as the evidence base changes. An additional learning point from the work completed is how methods used here could be altered in future work to optimise the findings and address limitations. In the final study, focus groups were used to explore experiences of engaging with the NON-STOP app. The pitfalls of this approach were discussed in detail in the discussion in Chapter six, however, to link this to the learning opportunity for the researcher, it is important to consider the potential changes for future work. Focus groups within the feasibility study were attended by children and families that engaged with the intervention. The findings therefore reflect a sub-group of the sample that engaged, and do not reflect the population. i.e., those who did not engage, did not have the opportunity to express

why. On reflection, this would need addressing in future intervention development and testing studies, with different approaches (discussed in Chapter six) selected. Relating back to the aims and objectives, the method still effectively addressed those, however, focus groups to inform refinement of interventions may not be optimal in the wider context of intervention design. This would need addressing in order to optimise engagement within the desired group, and maximise longevity of the intervention.

7.5 Key points for discussion

In each of the empirical chapters that present the studies completed and the development of the NON-STOP app, there are discussion points that relate to that particular piece of work. In this section, two discussion points have been selected to revisit in more detail. The topics are relevant when considering this project as a whole, and considering its impact on clinical practice and how it has produced original contributions to the literature. Given the lack of literature relating to the non-surgical treatment of Perthes' Disease, much of this contribution is given with reference to the development of digital self-management interventions. The discussion then moves to the key points for consideration and discussion for implementation of the NON-STOP app in the clinical field, and the challenges that this includes. Then the researcher presents a section discussing implementation of digital health interventions. Finally, self-management as a phenomenon is explored. With a particular focus on the notion of self-management in children's physiotherapy.

7.5.1 Self-management for Perthes' Disease

In recent years, there have been studies, including a meta-analysis from Panagioti et all, which outlined how self-management interventions can underpin significant positive outcomes for patients and health care services [288]. In this review, the authors were able to demonstrate that patient outcomes were maintained with self-management interventions whilst reducing the demand on the health care services.

Whilst this particular study was focused on adult care, similar results have been observed in child-health research involving children with physical disabilities [289]. Lindsay et al were also able to demonstrate improved outcomes including quality of life and reduced hospital visits.

In the context of this project to develop a digital self-management intervention, the need was demonstrated by the researcher in a case review which demonstrated a variation of care in the UK [28]. In this study, centres in the UK reported variation in many aspects of care including physiotherapy, surgical intervention and activity modification advice. In chapter three, children with Perthes' Disease and their families explained that the variation of care had impacts on their quality of life, and that there was a strong desire for education about the condition as well as input from relevant healthcare professionals. Most importantly, the participants expressed that the NON-STOP app was a suitable solution to deliver a self-management intervention.

Self-management offers many positive elements to those with health conditions, and in this project specifically, the use of psychological theory has been used to develop a self-management intervention. It is also possible to use the theoretical underpinning to explain why self-management has a positive impact. In this project, the SDT ensured that the motivating factors when creating the NON-STOP app related to three key components that lead to a sustained change in behaviour: autonomy, competence and relatedness [78]. In Chapter five, Table 5.3 – Key elements of the app and how they map to each of the theory elements, the specific elements of the NON-STOP in relation to SDT are discussed. The principle is the same when considering why self-management is positive for patients in healthcare. Providing the necessary tools to self-manage meets all three of the components of SDT. By educating to an adequate level, a patient will understand what to do and when, but also why they are being asked to self-manage. In this scenario, the patient is competent, because they have been educated to a level that means they can execute the self-management strategy, whatever that may be. They also have the ability to do it independently, demonstrating their autonomy. Finally, self-management interventions that have been developed using robust methods meet the needs of the user. Meaning that if done correctly, the patient will engage with a self-management

intervention that addresses their needs, demonstrating relatedness. There were examples of this in chapter six in the nested focus-groups where children who used the NON-STOP app and their families described the independence that the app had given them.

There is a lack of digital self-management interventions for children relating to physical activity or rehabilitation reported in the literature. In Chapter two, the work by Mannocci et al was discussed [73]. The umbrella review demonstrated that physical activity interventions of any type were mostly based in school settings and that further research should investigate in other environments. There have been digital behaviour change interventions developed in other child-health areas that have been reviewed in the literature, specifically one systematic review by Brigden et al in 2020 [290]. The authors identified studies which described effective digital interventions, reported promising interventions' characteristics, and described the user's experiences. The authors highlighted in the review that self-management skills demonstrated an increase in user knowledge, self-care, quality of life, symptom control and health service utilisation. Seventeen interventions were included in the review, seven were related to weight management, of which most included some measure of physical activity as an outcome. The interventions described by the authors had limitations, and of 17 interventions, only three were deemed 'quite promising' according to the criteria outlined. For an intervention to be deemed 'quite promising', there needed to be a demonstration of positive effect on the primary outcome, or a behaviour change outcome. Any interventions that did meet both outcomes were referred to as 'very promising', no interventions were identified as such. The majority of the interventions demonstrated high levels of bias and did not meet primary outcomes or behaviour change outcomes. Although 17 interventions were included in this review there is a lack of digital self-management interventions available in the rehabilitation space for children with health conditions. It is therefore difficult to truly map the findings of this systematic review to the findings of this doctoral project. Self-management of Perthes' Disease relies on engagement from all stakeholders, and future research is discussed later in this section, but should certainly include detailed evaluation.

Self-management and knowledge improvement has been a focus of gamification-style interventions for young people with chronic conditions and have demonstrated some positive results [291]. The findings of this study by Charlier et al are somewhat translatable to the NON-STOP app development, as they show promising findings relating to increased knowledge after using game-style interventions. Gamification was a significant part of developing the NON-STOP app, with participants of Chapter three explaining that any self-management intervention must be “fun, and like a game” but all stakeholders wanted increased levels of education. There are however relatively significant limitations to this review of interventions relating to the population. The degree to which self-management has been measured is mostly related to children with diabetes being able to effectively monitor their own blood-glucose levels [292]. Whilst a very important element of care for children with diabetes, not very relatable in the development of the NON-STOP app as monitoring is not a part of self-management. Overall, the review offers some reassurance to the methods of gamification for increasing knowledge for children with health conditions that could contribute to improved self-management.

Once again, there is clear alignment with the theoretical underpinning of this doctoral programme of work. Gamification has demonstrated improvement in motivation and adherence to desired behaviours in a range of physical and mental health conditions [293]. Linking this to SDT particularly, gamifying an intervention like the NON-STOP offers an opportunity for longer-term engagement through increasing levels of relatedness, competency and indeed autonomy increasing as the child’s ability level increases. “Levels and rewards” were discussed by children with Perthes’ Disease in the qualitative study. Rewards were integrated into the design and development, but levels were more difficult to incorporate given the testing period that was possible in a PhD. There are limitations to the gamification of interventions, one of which is overstimulation and in turn disengagement [293]. Fine balance is required, and further demonstrates the importance of key stakeholder/PPI input at key stages of intervention design and development.

As introduced above, self-management poses significant challenges, many of which were applicable to this project. To present all would be outside of the scope of this

project, the main challenges are similar to those in implementation of the NON-STOP intervention. Support systems are required for patients to engage with self-management interventions, for instance in the treatment of Perthes' Disease, the NON-STOP app was not designed to replace existing care. Any refined and implemented version of the NON-STOP app (post-definitive clinical trial) will be implemented to support existing care, reducing variation of care in the clinical setting. If not done with engagement from necessary systems such as input from clinical specialists, children with Perthes' Disease and their families are at risk of isolation, which carries much greater risk. Engagement with the intervention is another challenge with self-management interventions such as the NON-STOP app. Self-efficacy is a challenge in a self-management intervention, and was explored in the feasibility study for the NON-STOP app. Ensuring that users feel confident in using the intervention, and that it will lead to the desired outcome, i.e., the ability to self-manage, is key. Similarly, as seen in chapter six, any intervention must consider an appropriate dosage to prevent disengagement [285]. Once again, similar to implementation and the theoretical underpinning of the project, motivation is a factor and must be considered when aiming to increase self-management. With specific reference to digital health interventions, health literacy and accessibility cannot be ignored. These were discussed in Chapter five and have also been considered by the authors of the digital behaviour change interventions review above [290]. Digital interventions aimed at improving self-management often rely on a certain level of access, for example to a smart device and mobile data. Similarly, they rely on a baseline level of health literacy, and these must be considered, as they were in the development of the NON-STOP app. If any of these challenges are not considered, there is a risk that the desired population will become disengaged, and in turn isolated due to a lack of input and support [294]. This is a very serious issue given that self-management is aiming to establish a stronger connection to care provision.

Addressing the challenges discussed in the future development and integration of the NON-STOP app in clinical practice could be guided using methods described in the implementation section above. There is a strong overlap with the issues to address in

the implementation of the intervention and challenges of self-management. In a wider sense, the challenges that face the NON-STOP app are not exclusive to this digital self-management intervention. In the development of other digital self-management interventions for children, one must consider the key stakeholders and environments in which it may exist. Differences will exist in different patient populations, and this must be acknowledged in order to maximise engagement.

7.5.2 Future implementation of the NON-STOP app

Implementation science is a vital part of intervention development and evaluation, and provides methods and approaches to increase the uptake of an intervention in its intended area [295]. Applying an implementation theory to an intervention can be used to reduce the time between intervention development and it being adopted in clinical practice.

Arguably the biggest challenge facing the uptake of the NON-STOP app, a novel digital self-management intervention for the non-surgical treatment of Perthes' Disease, is implementation. There are many factors to consider when planning to incorporate the uptake of the NON-STOP app in current clinical practice. Some of these challenges are considered in detail in the limitations section below. Other challenges were considered in the development and early testing of the intervention as per available guidance [44]. For example, the integration of the views of the users/population for whom it was developed. In PPI and Project Advisory Group (PAG) meetings, children and families explained that access to apps was easier if mobile data/internet access was not required for use. Work with the developers was completed to ensure that users could complete their sessions and relevant updates to servers, etc. took place once internet connection was re-established. To address the true demands that complete implementation poses was outside of the scope of this PhD. It was, however, important within this thesis to consider certain aspects of future implementation relevant to the NON-STOP app.

Implementation of the NON-STOP app in the future must involve maximised engagement with key stakeholders. In the context of the NON-STOP app, this involves

children with Perthes' Disease and their families, and many aspects of engagement were evaluated in Chapter six. Elements of engagement that were not evaluated in the app-testing study were engagement with clinicians. In order to maximise uptake of this novel intervention to support self-management, careful consideration must be given to clinicians who care for children with Perthes' Disease. In the work prior to this doctoral programme of work, the researcher was able to demonstrate widespread variation in the UK [28]. In Chapter four, clinical consensus recommendations were produced, and the published work [219] as well as national dissemination to clinicians potentially reduced the variation of care. To sustain uptake of the NON-STOP and in turn, maximise a successful implementation, there needs to be clear engagement with clinicians who care for children with Perthes' Disease and their families. To do this, similar theories and approaches to those used in this project could be employed, for example, the use of psychological theories relating to behaviour change such as SDT and SEM [78, 109]**.** The majority of the uptake and implementation of the NON-STOP app in a 'real world' clinical setting would involve engagement with physiotherapists and surgeons working in children's orthopaedics. Nationally the researcher has developed a strong relationship in this space through the work completed in this project. One example is that the researcher currently chairs an Allied Health Professionals and nursing committee within BSCOS. Utilising these networks to develop relationships with relevant teams aligns with the guidance of the MRC Framework which encourages users to engage with relevant stakeholders as part of the implementation of an intervention [44].

Another practical consideration regarding the implementation of the NON-STOP app due to it being a digital intervention was where the app would be hosted. In the context of the NON-STOP app, this means where the app would be placed, for the users to be able to access it, such as a webpage that clinicians can direct potential users to in order for them to download the app. It also relates to regular maintenance and software updates that are not substantial. For instance, to ensure that apps continue to function with the most current operating system of the device. For the purpose of this doctoral programme of work, and a short period after project, funding was secured for the developers (HMA) to host the NON-STOP app on their servers.

Longer term there needs to be a plan regarding hosting, not only including the securing of funding to do so. Prior to 2021, there would be the potential for the NON-STOP app to be adopted by the NHS app library, however this was decommissioned in 2021 [296]. Applications such as the NON-STOP app are still selected by clinical policy teams, and recommended on the NHS website at relevant sub-pages. It would be reasonable for future implementation work to include a decision making process in which a suitable host is selected. Hosting somewhere that is well known to the clinical population like the NHS website would be sensible, and would align with common aims of implementation theories which are to reduce research waste [297]. Alternative locations were conceivable; they include the potential to have the NON-STOP app hosted on another relevant webpage that would result in acceptable levels of engagement. One example would be the STEPS charity website, the researcher, as well as members of the supervisory team have developed strong relationships with the charity and having the app hosted on that page would also be reasonable.

With all of the options presented, one must recognise one of the most significant implications that come with digital health technologies, and that is cost. As operating systems develop and update, so must applications that are used on them. Updates come at a cost, and in the context of the NON-STOP app, this would likely include liaison with NHS England to adopt the technology and approve it for recommendation on the NHS website. Correct at the time of writing this thesis (October 2024), the most common way of assessing a digital technology for uptake within the Health and Social care domain, was using a Digital Technology Assessment Criteria (DTAC) [298]. The DTAC is a collection of criteria that can be used by organisations at the procurement stage to ascertain whether a digital technology meet a set of minimum baseline standards [299]. This work has been designed with reference to one framework that has been used in this doctoral programme of work, the NICE ESF [223]. As a result, the criteria assess many of the elements of intervention development that have been employed in this project, including usability and acceptability.

7.5.2.1 Implementation approaches

There are a number of theories/approaches that could be applied to the NON-STOP app in order to evaluate the longer term implementation of the intervention. One option would have been to apply the RE-AIM framework to the development of the NON-STOP intervention [300]. RE-AIM is a tool used to evaluate the potential impact of an intervention in its intended population [301]. The evaluation is based on five components: Reach, Effectiveness, Adoption, Implementation and Maintenance (RE-AIM). Put into context for the NON-STOP app, applying this framework would allow the researcher to systematically evaluate its real world impact, ensuring it serves its intended population, i.e. children with Perthes' Disease and their families. As well as this, it would allow the researcher to evaluate its impact on clinicians, and ensure it maintains long-term engagement.

RE-AIM has been used in child-health, albeit not in used for digital health interventions, to assess implementation of interventions that are somewhat similar to the NON-STOP app. In 2023 Briatico et al used the RE-AIM framework to evaluate a parent-focused intervention targeting childhood obesity [301]. The intervention was based on behaviour change techniques, including education sessions that children and parents attended at three stages in the intervention period (13-weeks). The intervention was well defined, with a previously published pilot study [302] outlining the intervention and a further report discussing the findings of the pilot study [303]. The authors provided a clear outline of the RE-AIM dimensions and practical examples of what measures were collected and when. Using the RE-AIM dimensions, the authors were able to demonstrate that they successfully reached their intended participants, children with obesity. Effectiveness was demonstrated with weight loss over the intervention period. A key limitation of the use of RE-AIM in this study is that the authors did not factor in any review of long-term maintenance data due to the study focusing on the implementation of the intervention which was a 13-week programme. The lack of long-term monitoring in this study is not uncommon and a key characteristic of RE-AIM, which is resource intensive, requiring data collection on each dimension for a long period of time. With recent literature regarding RE-AIM suggesting that maintenance should be measured around one to

two years post initial implementation [304]. The robust nature of the implementation framework does however mean that the strength of its application is that you produce a comprehensive assessment of the implementation of the intervention. Applying this method to the implementation of the NON-STOP app would be appropriate. It would allow the researcher to gain an insight into important elements of the intervention, for example, areas that are well engaged with, or clinical measures that are reviewed after a period of use. For Perthes' Disease it would include the use of the measures included in the core outcome set. It would need consideration and reasonable support both from expertise and resources in order to measure all five dimensions.

Another approach that would be possible in the implementation of the NON-STOP app would be to apply a COM-B model [305]. The COM-B model focuses on three main components: Capability, Opportunity and Motivation, and relates these to Behaviour. This concept has been discussed in Chapter five however this was mostly aimed at the development of the intervention rather than any focus on implementation. It would be particularly appropriate to consider this given the focus on the intended change in behaviour from using the NON-STOP app. The COM-B model has been applied to interventions that focus on physical activity in young people [306]. Here the authors were able to use the COM-B to design and evaluate the intervention and focus on which components in particular affected the change in behaviour. As discussed, this would be appropriate to apply in a more detailed sense to the NON-STOP app to allow for evaluation of how any sustained change in behaviour was achieved. It is, however, worth noting that when compared with an approach like RE-AIM, the COM-B model would not provide as comprehensive of an evaluation. For instance, whilst it would identify factors affecting behaviour that relate to the user, it does not consider broader factors that influence change in behaviour. An example in relation to the NON-STOP app would be that it would not consider the implications of environmental support systems like increased and sustained engagement from clinicians.

Selecting an implementation theory or approach should be done with careful consideration, and in 2018 Birken et al developed a tool to assist in the comparison

of theories and approaches. It is called the Theory, Model and Framework Comparison and Selection Tool (T-CaST) [307, 308]. The authors created this tool which allows researchers to compare theories, models and frameworks by applying certain criteria that relate to four commonly used domains: usability, testability, applicability and acceptability. Two of these measures were the focus of chapter six of this doctoral programme of work. More than allowing researchers to compare and select the implementation approach, the authors also explained how T-CaST has the potential to improve the reporting of criteria used when selecting an implementation approach.

Taking in to account the above, the implementation of the NON-STOP app as a digital health intervention will require specific expertise to facilitate robust evaluation in order to ensure an effective adoption and sustained use in clinical practice. Engagement with key stakeholders including children with Perthes' Disease, their families and clinicians who care for them will be paramount. As outlined above, there are stakeholders that were not wholly considered in the development of the intervention that would need to be included, such as organisations who may host the intervention and certainly those who will fund the ongoing technological support of the digital intervention. Future research should include the integration of a proven framework to aid implementation; two potential options have been provided here. It would also be useful to use a decision making tool such as presented above in T-CaST [307, 308]. The use of a robust framework will ensure that the NON-STOP app meets the needs of environment in which it is used as well as the children and families that engage with it. This will maximise engagement in the long term and lead to a sustained change in behaviour which would be optimised self-management of Perthes' Disease.

7.6 Strengths and limitations

In each of the relevant chapters, the strengths and limitations of each study/element of the project have been discussed. The following section summarises the key strengths and limitations of the overall project. Where possible, relevant literature surrounding the topic has been considered.

7.6.1 Development of evidence-based digital intervention

App-development and use in every element of life has become common practice, as of August 2024, there are over two million apps available on the Apple App Store, and almost 2.4 million apps on the Android Play Store [309]. App development and use in healthcare is also rising, with a multitude of apps for children in the healthcare and education space. Many of which were discussed in Chapter five. As outlined in chapter five, there are methodologies that exist for the development of complex interventions [138]. There are, however, no robust methodologies that guide development of digital self-management interventions for children with a focus on physical activity. Nevertheless, complex interventions are common practice in health research, particularly in studies where rehabilitation, physical activity and self-management are involved [310-312].

Taking the above into consideration, a key strength of this doctoral programme of work is that the NON-STOP app was developed using methods that are becoming relatively common practice in studies that are funded by influential organisations including the National Institute for Health and Care Research (NIHR). The application of the MRC framework and NICE Evidence Standards Framework (ESF), as well as several behavioural psychological theories highlight the rigorous approach that was taken in this project [44, 78, 109, 145, 165, 223]. A mix of methods were employed across the project, in the studies outlined in section 7.2. These studies provided novel findings in the field of Perthes' Disease and allowed the researcher to produce the content necessary to apply the blended intervention development approach.

Paying particular attention to the MRC Framework and NICE ESF [44, 223], it is possible to suggest that future research could explore a reconciliation between these two pieces of work to provide a clear guidance for those developing digital interventions. This would most likely take the form of a new document; however, the potential impact should not be ignored given its potential application across a number of disciplines. Particularly as the health, and healthcare research world continues to become more and more digital.

7.6.2 Methodological approaches taken in the project

A factor that posed a significant challenge in this doctoral programme of work was the number of methodologies that were utilised. Qualitative methods, Delphi methodology and mixed-methodology were all new experiences for the researcher, and required input and support from those with experience in each. There was an implementation of the skills learned over the course of the project, for example, lessons learned from experience and reflection in the qualitative study in chapter three allowed for a more efficient data analysis in chapter six. Ultimately, the methods applied were chosen to ensure that the relevant stakeholders were integrated in the research design, and in turn the findings most applicable to them.

It would be unrealistic to expect a doctoral researcher to complete this alone, and with expertise from the supervisory team, and sometimes wider (such as qualitative support from SP in chapters three and six), each study was successfully completed. That being said, as a result of this programme of work, the researcher has now developed at least a foundation level of understanding of a mix of methodologies. These methodologies can be employed when developing an intervention, which will likely be a significant part of the researcher's future as a clinical academic children's physiotherapist. It will also give the researcher the foundation to develop support mechanisms for future healthcare researchers. As well as learning to carry out these methodologies with support, it has also been possible to reflect on the time management skills that have been developed as a result of using a mix of methods.

Understanding logistical implications around conducting studies that require ethical approval from NHS REC/HRA due to the involvement of NHS patients.

Applying the research methods used in this thesis has been a challenge, not least given the added complexities that child-health research introduces. In chapters 3 and 6, children were involved as participants, as young as five years old in the studies. To the knowledge of the researcher, there is no guidance on how to tailor the research methods to children. The application of this mix of methods, does align to the pragmatic approach that has been employed throughout the thesis. One example is the utilisation of rapport building periods within the online-conducted interviews with children in Chapter three. Developmental appropriateness is something to be considered for each participant in child-health research. Language skills, non-verbal communication, and monitoring levels of engagement are particular challenges of qualitative research with children. To give a quantitative example of a challenge, when collecting app-testing data in Chapter six, it may be reasonable to consider a certain level of engagement 'acceptable' for a younger child compared to an older child, or indeed vice versa, but there is no available guidance for this. Once again, a pragmatic approach was applied with regular PPI and PAG input. This approach would be recommended in the absence of any robust guidance.

It would have been possible to apply a mixed-methods approach to the whole project as opposed to a mix of methods. Recent research has demonstrated the positive impact of employing mixed-methods in intervention studies in children and adolescents [313]. In this methodological review, Fabregues et al reviewed mixed-methods in intervention studies in emotional and behavioural disorders. Whilst the area of practice is not related to physical activity or rehabilitation the authors summarised the findings of the studies and reported that in many of the included studies, there were reports of positive impact of mixed-methods. Included in the positive impact was elements such as more contextualisation of findings, similar to the positive impact of applying mixed-methods in Chapter six.

There are a number of factors that were considered in the decision-making process when selecting the methodology. The most important factor was that each method

chose, aligned with the views or aims of the researcher. This project has been completed with a pragmatic approach, with methods selected to meet the aims and objectives to deliver a usable and acceptable digital self-management intervention. The methods were agreed upon by not only the researcher, but with input from a supervisory team. The wider team included complex intervention development and evaluation specialists, qualitative methods specialists and clinical academic leaders that have substantial experience of develop clinical consensus. The methods chosen ensured the aims and objectives of each study were met, and in turn, the aims and objectives of the overall project were met.

7.6.3 PPI input

Prior to this doctoral project, PPI and input from children with Perthes' Disease advised the researcher that research should take place to develop and test an app. Originally, there was an idea to develop a self-management intervention in the form of a website. Children unanimously explained that an app would better support them to complete their physiotherapy exercises. It is well described in the literature and outlined in the mission statement of the NIHR that PPI are paramount in health and care research [314]. In the most recent survey by the NIHR, children, including those in the 0-6 years old category, reported that they felt valued to be involved in research. Strong PPI is also an important part of intervention development [44]. From the outset of the PhD, a project advisory group (PAG) was established which included children with Perthes' Disease, family members and clinicians. The PAG was chaired by a member of the public, who also held the role of General Manager of the children's charity STEPS Worldwide. The group was maintained throughout the duration of the PhD and has had input in many different aspects of the project. To highlight some of the key events:

- Design of research study materials such as participant information sheets and topic guides
- Design of the study website, hosted by the researcher's clinical employers

- Dissemination of study findings in various forms such as online videos and graphics
- Cartoon design including the ‘bringing-to-life’ of Bobby the Bone and other characters in the NON-STOP app

The PPI and PAG activity throughout the project aligned with the theoretical underpinning of the project which focus on factors that motivate people to change their behaviour, and hopefully, sustain that change in behaviour [44, 78, 109, 145, 223]. It also very much aligns with the pragmatic approach of the researcher. To try and better understand how to optimise non-surgical treatment of Perthes’ Disease and develop an acceptable and usable digital self-management intervention, it seemed only too reasonable to include children with Perthes’ Disease, their family members and the clinicians who care for them in the design and delivery of the project. The success of this project is attributable to the PAG and PPI members that have contributed to this project. Future work regarding the NON-STOP app, as well as other clinical research conducted by the researcher, will continue to have PPI as a central role in the research process.

PPI has, understandably, become such a significant focus of organisations such as the NIHR, it would be possible to evaluate the PPI impact from this study using something like the Public Involvement Impact Assessment Framework (PiiAF) [315]. The PiiAF is a framework that can be used to assess the impact of involving members of the public in their research, and has been done in settings such as mental health research [316]. There is now international guidance available to assist researchers in the reporting of PPI in research [317]. Realistically, any formal evaluation of PPI impact was outside of the scope of this project. In an attempt to maintain an effective oversight of what had been discussed in PPI activities from the start of the project (2021), the researcher kept a PPI log. These were stored in a variety of forms, audio notes reflecting on PPI sessions were taken after sessions and written reflections were produced to outline how PPI activities influenced the work in this project.

7.6.4 Potential implementation of the NON-STOP app

Understanding the necessary steps to bringing evidence-based interventions from research in to the 'real world' is vital [318]. Implementation science plays a key role in ensuring that interventions are not only effective in controlled settings but also scalable and sustainable in practice [44]. Implementation science helps identify barriers and enablers that could affect the uptake and sustainability of complex interventions, which is essential in projects such as this [319, 320]. In this project, it was not possible to fully address barriers and/or enablers to implementation after preliminary testing.

Implementation was considered throughout the project as part of the programme theory, displayed using logic models (Appendix I). These models outlined potential routes to implementation, despite a formal strategy being outside of the scope of this project. When considering larger scale implementation of an intervention like the NON-STOP app, a multitude of factors must be addressed, ranging from clinical integration to sustained engagement. These considerations apply not only to the NON-STOP app, but also to the broader theory and approaches available when considering implementation of an intervention.

In the development of the NON-STOP app, key stakeholders were involved through PPI activities and inclusion in the studies. While a comprehensive implementation strategy was not developed as part of this doctoral project, ensuring a user-centred design increases the feasibility of future real-world implementation. By involving children with Perthes' Disease, their families and clinicians in Chapter three, and a diverse sample of clinicians in Chapter four, the researcher ensured the NON-STOP app was grounded in the needs of both the clinical population and its intended users. In Chapter six, some degree of implementation is evaluated, with engagement with the NON-STOP app and factors that affected impact and uptake of the intervention. However, a more extensive evaluation is required to fully assess the implementation of the NON-STOP app within its intended real-world context. This would need to begin with evaluation of the intervention in terms of efficacy and comparison to what

has been delivered in practice. More details regarding the practical challenges of evaluation are discussed in section 7.8.

7.7 Impact on practice and policy

In each of the studies throughout this project, the implications for practice and future research were discussed. In this section the researcher has provided an overview of the impact of this doctoral project in relation to policy and practice. The impact relates to clinical practice, including relevance to both children's orthopaedic physiotherapy and surgery, as well as the wider clinical research context.

7.7.1 Impact in a clinical context

Through the objectives set out in this doctoral fellowship, work has been completed which independently has positively impacted the understanding that we have about Perthes' Disease. The standout piece of work relating to clinical impact is the clinical consensus recommendations for the non-surgical treatment of Perthes' Disease which supports the practice of children's orthopaedic specialists including physiotherapists, clinical nurse specialists and surgeons [219]. In the absence of robust evidence, these clinical consensus recommendations are as close to agreement that the children's orthopaedic community has come in regard to non-surgical treatment. Successful dissemination through various methods, such as social media, has resulted in nationwide interest in the findings. The clinical consensus study in Chapter four was selected for a focus review by the Bone and Joint Journal, and selected as 'Article of the month' by the same journal [321]. Presentations at national conferences such as the British Orthopaedic Association and BSCOS annual conferences have increased the impact of the findings in the clinical context. There has also been dissemination of materials such as the consensus recommendations infographics to over 40 departments in the UK, including children's orthopaedic specialists, but also wider members of the MDT such as Paediatric Accident and Emergency colleagues, community paediatricians and GPs. In the patient population,

there has been strong engagement after sharing the findings with children and families through charity outputs and webinars.

The findings of this study in the context of the doctoral programme were vital in developing the clinical content for the NON-STOP app and were integrated as described in Chapter five. It is hoped that the findings, aided by the wide dissemination, will result in a reduction in the variation of care. In the future, this will ensure that regardless of the geographical location of a child with Perthes' Disease, the care they receive will be based on the best available evidence.

7.7.2 Impact in children's orthopaedic research

Clinical trials in children's orthopaedics have made significant advances in the last ten years, with a development of numerous successful funding calls to deliver robust, randomised clinical trials in a number of conditions [322]. Nevertheless, many questions remain to be answered, including several that involve a strong physiotherapy influence and are related to self-management and/or rehabilitation. The first children's orthopaedic trials addressing rehabilitation are now underway, with the ROBUST [323] and SPELL [324] trial seeking to evaluate the effectiveness of strengthening and stretching, respectively, for children with cerebral palsy.

Whilst the aim of this doctoral project was to deliver and begin testing of the NON-STOP app, the doctorate has provided an opportunity for the researcher to establish a profile as a clinical academic in children's orthopaedics. Nationally, allied health professionals are under-represented in clinical academia and providing support for these professions is a focus of the NIHR, as set out in their strategy document "Best Research for Best Health: The Next Chapter" [325]. Throughout the duration of the doctorate, the researcher has had the opportunity to share learning and skills that have been developed with wider members of the children's orthopaedic community, but also the allied health professions and early career researcher groups within organisations such as the NIHR. In the future there is a need for greater capacity development in this space to support further advances in the children's orthopaedic physiotherapy field.

The findings from this programme of work have been shared with the clinical community, elements of the thesis have recently been included in a scoping review by Beni et al [326]. In the scoping review, the authors highlight the qualitative research from Chapter three [155] as well as the previously completed systematic review [34]. The authors have included a focus on the findings from the clinical consensus study to provide a clinical pathway for initial management [219].

Over the course of this doctoral programme of work, an intervention has been that is usable and acceptable has been produced. It has been refined, and is prepared for integration in a randomised clinical trial comparing surgical and non-surgical treatment of Perthes' Disease (The Op NON-STOP study), which is discussed below.

7.8 Future research

Throughout this thesis, there has been regular reference through the aims and objectives to a future "definitive clinical trial". Therefore, a reasonable next step would be to compare the NON-STOP self-management intervention to existing care. These methods are regularly utilised in clinical research, and particularly in rehabilitation studies where novel interventions are compared with existing care [327]. An example was used when comparing physiotherapy alone with additional "eHealth", which was a digital intervention including self-management materials for patients with spinal pain. The materials included education around pain management and behaviour change techniques to help patients understand their pain and manage the condition. The intervention was well described and used TIDieR guidelines [63] to report the intervention and included training of the physiotherapists delivering the experimental intervention (eHealth). The control intervention (usual care) is also well defined, with national practice guidelines for spinal pain described. The methods used to evaluate the effectiveness of this novel intervention compared to the control intervention are well defined and appropriate.

Regarding the evaluation of digital interventions, guidance is available which emphasises the importance of conducting randomised clinical trials to provide definitive evidence on the effectiveness of these interventions [328].

Recommendations from Murray et al were based on the findings of an international consensus workshop and stipulate that a trial is needed, however this should only be done once certain criteria are met. One is that the intervention is “stable” i.e. any future developments are minor, in the context of the NON-STOP app, the content has been well defined, and the delivery systems are in place, refinement would include making the intervention applicable for longer term use rather than a shorter testing period. The remaining criteria relate to the intervention having high fidelity and a potential for meaningful benefits in the clinical sense. Fidelity has been examined somewhat in this project, but not with a specific approach to demonstrate fidelity, rather to demonstrate aspects of it such as acceptability and usability [329].

The key limitation of applying this methodology to the Perthes’ Disease population is that usual care has never previously been standardised, as demonstrated in the case review and systematic review completed by the researcher [28, 34]. Any future study that compared the NON-STOP to ‘usual care’ would therefore be difficult to accurately evaluate due to the variation in what would be the control arm.

7.8.1 The Op NON-STOP study

At the point of submission of this doctoral thesis (December 2024), refinement of the NON-STOP app has taken place based on the findings of chapter six, and engagement with PPI members. As discussed in Chapter one, there has been a strong desire from the clinical community, as well as the patient population, to compare surgical and non-surgical treatment of Perthes’ Disease. The best way to provide an answer as to how clinicians should best treat children with Perthes’ Disease is a randomised clinical trial. In 2024, the researcher, along with two members of the supervisory team (DP and DK) were involved in the successful application of a NIHR Health Technology Assessment (HTA) grant to compare surgical and non-surgical treatment of Perthes’ Disease (ID: NIHR152309, ISRCTN83315571) [330, 331]. This multi-centre, prospective, randomised superiority trial children will be randomly allocated to receive surgical, or non-surgical intervention, described in a publication authored by the researcher [332]. If randomised to the non-surgical arm of the study, they will

receive a one-off best evidence advice session with a trial trained physiotherapist. The content of this session has been designed based on the findings of this doctoral programme of work. The participants in the non-surgical arm will also be given access to self-management materials, i.e. a refined version of the NON-STOP app. The study has been designed with a pilot phase in the first 12-months, and throughout the course of the trial, there is the opportunity to embed elements of the implementation processes discussed in section 7.5.1. Elements of implementation that will be evaluated are the level of engagement and the effect of greater engagement (i.e. more app use) and the outcomes included in the trial. These outcomes are the same that were used in chapter six, and are based on the core outcome set for Perthes' Disease [95]. There are some limitations regarding the NON-STOP app being used as part of a comparator in a clinical trial. For instance, one could argue that the NON-STOP app has not undergone effectiveness/efficacy testing that aligns with best practice for interventions [44]. It was not the researcher's original plan to integrate directly from this doctorate in to a clinical trial. The initial plan would have been evaluation of the NON-STOP app plus 'usual care' compared with usual care. However, the choice was made amongst intervention specialists and those with expertise in conducting clinical trials in children's orthopaedics to move forward with the clinical trial with the findings from this doctoral programme of work as a key component of the non-surgical intervention. The findings have helped to standardise the non-surgical management, as described, to support the non-surgical intervention within the trial which includes the NON-STOP app supplementing usual care. Given the degree of variation that has been described in 'usual care', this project has served as effective in providing evidence to optimise the non-surgical treatment of Perthes' Disease.

Considering future research in a wider sense and factoring in the direction of healthcare with regards to digital health interventions, it is likely that the findings of this project could be used to conduct similar research in other children's orthopaedic conditions. For instance, as discussed, the methods of intervention development used in this project could be applicable to other conditions where a digital self-management intervention is worthy of exploration.

7.9 Conclusion

The aim of this doctoral programme of work was to utilise the experiences and recommendations of key stakeholders, to deliver a digital self-management intervention for the non-surgical treatment of Perthes' Disease. The aim was then to assess the usability of the tool, and the acceptability in preparation for a future definitive clinical trial. This aim was achieved using a mix of methods, each applied to meet the specific aims and objectives of the project. The overarching theories (SDT and SEM) that underpinned the project, and the pragmatic approach of the researcher, contributed to the objectives being met. PPI and key stakeholder involvement was imperative in this thesis. Their input at key milestones ensured a deeper understanding of factors that optimised engagement with the intervention. This doctoral programme of work has the potential to be used as an example of methods that could be applied to other areas of children's orthopaedics or rehabilitation where self-management interventions are a potentially useful direction for clinical care.

The findings of this project have contributed in an original manner to developing the understanding of Perthes' Disease in a number of ways. In chapter three, the experiences and understanding of children with Perthes' Disease, their families and the clinicians who care for them were captured. This had not previously been done and has contributed a level of understanding to the scientific community, whilst providing valuable information in the development of the NON-STOP app. In chapter four, clinical consensus recommendations provided guidance for clinicians regarding non-surgical treatment of Perthes' Disease for the first time in this patient population. In chapter six, the findings from a mixed-methods study suggest that the NON-STOP app is usable and acceptable to children with Perthes' Disease and their families. Further work is required to further evaluate the NON-STOP app in real world settings, and the inclusion of key stakeholders and PPI members will be vital.

Using the methods described, this doctoral programme of work has produced novel findings that contribute to the optimisation of the non-surgical treatment of Perthes' Disease. These include clinical consensus recommendations that have the potential

to reduce variation in care, which children with Perthes' Disease and their families expressed as an important issue in previous experiences. Based on these findings, the NON-STOP app was created and in preliminary testing, yielded a very promising response from uses who found it to be acceptable and usable. The findings from this thesis are currently integrated in the first prospective, randomised clinical trial to take place for children with Perthes' Disease which looks to address one of the most important topics in children's orthopaedic research. A comparison on surgical containment and optimised non-surgical treatment of Perthes' Disease.

References

1. Calvé, J., *Sur une forme particulière de pseudo-coxalgie greffée sur des déformations caractéristiques de l'extrémité supérieure du fémur.* Rev Chir, 1910. **42**: p. 54-84.
2. Legg, A.T., *An obscure affection of the hip-joint.* The Boston Medical and Surgical Journal, 1910. **162**(7): p. 202-204.
3. Perthes, G., *Über Arthritis deformans juvenilis.* Deutsche Zeitschrift für Chirurgie, 1910. **107**: p. 111-159.
4. Joseph, B., H. Shah, and D.C. Perry, *Epidemiology, natural evolution, pathogenesis, clinical spectrum, and management of Legg-Calvé-Perthes.* J Child Orthop, 2023. **17**(5): p. 385-403.
5. Kim, H.K.W. and J.A. Herring, *Pathophysiology, Classifications, and Natural History of Perthes Disease.* Orthopedic Clinics, 2011. **42**(3): p. 285-295.
6. Waldenström, H. and I. GOLDIE, *The First Stages of Coxa Plana.* Clinical Orthopaedics and Related Research®, 1984. **191**: p. 4-7.
7. Joseph, B., G. Varghese, K. Mulpuri, N. Rao K. L., and N.S. Nair, *Natural Evolution of Perthes Disease: A Study of 610 Children Under 12 Years of Age at Disease Onset.* Journal of Pediatric Orthopaedics, 2003. **23**(5): p. 590-600.
8. Hyman, J.E., E.P. Trupia, M.L. Wright, H. Matsumoto, C.-H. Jo, K. Mulpuri, et al., *Interobserver and Intraobserver Reliability of the Modified Waldenström Classification System for Staging of Legg-Calvé-Perthes Disease.* JBJS, 2015. **97**(8): p. 643-650.
9. Varela-García, M., C. Torrijos-Pulpón, L. Pino-López, Y. Farrag, R. Forneiro-Pérez, J. Conde-Aranda, et al., *A retrospective study describing the acetabular consequences of Legg- Calve-Perthes disease.* BMC Musculoskeletal Disorders, 2024. **25**(1): p. 753.
10. Melin, L., Z. Rendek, and Y.D. Hailer, *Recommendations for physiotherapy and physical activity for children with Legg-Calvé-Perthes disease: a survey of pediatric orthopedic surgeons and physiotherapists in Sweden.* Acta Orthop, 2023. **94**: p. 432-437.

11. Rosenberg, A.R., L. Orellana, C. Ullrich, T. Kang, J.R. Geyer, C. Feudtner, et al., *Quality of Life in Children With Advanced Cancer: A Report From the PediQUEST Study.* J Pain Symptom Manage, 2016. **52**(2): p. 243-53.
12. Kim, H.K., K.D. Wiesman, V. Kulkarni, J. Burgess, E. Chen, C. Brabham, et al., *Perfusion MRI in Early Stage of Legg-Calvé-Perthes Disease to Predict Lateral Pillar Involvement: A Preliminary Study.* J Bone Joint Surg Am, 2014. **96**(14): p. 1152-1160.
13. Stulberg, S.D., D.R. Cooperman, and R. Wallensten, *The natural history of Legg-Calvé-Perthes disease.* JBJS, 1981. **63**(7): p. 1095-1108.
14. Perry, D.C., C.E. Bruce, D. Pope, P. Dangerfield, M.J. Platt, and A.J. Hall, *Legg-Calvé-Perthes disease in the UK: geographic and temporal trends in incidence reflecting differences in degree of deprivation in childhood.* Arthritis Rheum, 2012. **64**(5): p. 1673-9.
15. Perry, D.C., D.M. Machin, D. Pope, C.E. Bruce, P. Dangerfield, M.J. Platt, et al., *Racial and geographic factors in the incidence of Legg-Calvé-Perthes' disease: a systematic review.* Am J Epidemiol, 2012. **175**(3): p. 159-66.
16. Hall, A.J., D.J. Barker, P.H. Dangerfield, and J.F. Taylor, *Perthes' disease of the hip in Liverpool.* Br Med J (Clin Res Ed), 1983. **287**(6407): p. 1757-9.
17. Bahmanyar, S., S.M. Montgomery, R.J. Weiss, and A. Ekbom, *Maternal smoking during pregnancy, other prenatal and perinatal factors, and the risk of Legg-Calvé-Perthes disease.* Pediatrics, 2008. **122**(2): p. e459-64.
18. Perry, D.C., C. Thomson, D. Pope, C.E. Bruce, and M.J. Platt, *A case control study to determine the association between Perthes' disease and the recalled use of tobacco during pregnancy, and biological markers of current tobacco smoke exposure.* Bone Joint J, 2017. **99-b**(8): p. 1102-1108.
19. Lindblad, M., A. Josefsson, M. Bladh, G. Sydsjö, and T. Johansson, *Risk factors during pregnancy and delivery for the development of Perthes' disease, a nationwide Swedish study of 2.1 million individuals.* BMC Pregnancy Childbirth, 2020. **20**(1): p. 192.

20. Wiig, O., T. Terjesen, S. Svenningsen, and S.A. Lie, *The epidemiology and aetiology of Perthes' disease in Norway. A nationwide study of 425 patients.* J Bone Joint Surg Br, 2006. **88**(9): p. 1217-23.
21. Burwell, R.G., P.H. Dangerfield, D.J. Hall, C.L. Vernon, and M.H. Harrison, *Perthes' disease. An anthropometric study revealing impaired and disproportionate growth.* J Bone Joint Surg Br, 1978. **60-b**(4): p. 461-77.
22. Hall, A.J., D.J. Barker, P.H. Dangerfield, C. Osmond, and J.F. Taylor, *Small feet and Perthes' disease. A survey in Liverpool.* J Bone Joint Surg Br, 1988. **70**(4): p. 611-3.
23. Hailer, Y.D. and O. Nilsson, *Legg-Calvé-Perthes disease and the risk of ADHD, depression, and mortality.* Acta Orthop, 2014. **85**(5): p. 501-5.
24. Perry, D.C., D. Pope, C.E. Bruce, P. Dangerfield, A.J. Hall, and M.J. Platt, *Hyperactivity and the psychological burden of Perthes disease: a case-control study.* J Pediatr Orthop, 2013. **33**(6): p. 644-9.
25. Perry, D.C., B. Arch, D. Appelbe, P. Francis, J. Craven, F.P. Monsell, et al., *The British Orthopaedic Surgery Surveillance study: Perthes' disease.* The Bone & Joint Journal, 2022. **104-B**(4): p. 510-518.
26. Annamalai, S.K., R. Buckingham, and J. Cashman, *Perthes disease: a survey of management amongst members of the British Society for Children's Orthopaedic Surgery (BSCOS).* J Child Orthop, 2007. **1**(2): p. 107-13.
27. Do, D.H., M.F. McGuire, C.H. Jo, and H.K.W. Kim, *Weightbearing and Activity Restriction Treatments and Quality of Life in Patients with Perthes Disease.* Clin Orthop Relat Res, 2021. **479**(6): p. 1360-1370.
28. Galloway, A.M., C. Holton, V. Parnami, M. Wood, J. Craven, N. Green, et al., *A case review to describe variation in care following diagnosis of Perthes' disease.* 2020. **1**(11): p. 691-695.
29. Herring, J.A., H.T. Kim, and R. Browne, *Legg-Calve-Perthes disease. Part II: Prospective multicenter study of the effect of treatment on outcome.* J Bone Joint Surg Am, 2004. **86**(10): p. 2121-34.

30. Wiig, O., T. Terjesen, and S. Svenningsen, *Prognostic factors and outcome of treatment in Perthes' disease: a prospective study of 368 patients with five-year follow-up.* J Bone Joint Surg Br, 2008. **90**(10): p. 1364-71.

31. Alliance, J.L. *What are the short term and long term outcome of surgery compared to non-surgical care in the treatment of Perthes Disease? (JLA PSP Priority 4)*. 2018 [cited 2024 23/10/2024]; Available from: https://www.jla.nihr.ac.uk/priority-setting-partnerships/paediatric-lower-limb-surgery/priority-4-from-the-paediatric-lower-limb-surgery-psp.htm.

32. Vella-Baldacchino, M., D.C. Perry, A. Roposch, N. Nicolaou, S. Cooke, P. Ellis, et al., *Research priorities in children requiring elective surgery for conditions affecting the lower limbs: a James Lind Alliance Priority Setting Partnership.* BMJ Open, 2019. **9**(12): p. e033233.

33. Perry, D.C., J.G. Wright, S. Cooke, A. Roposch, M.S. Gaston, N. Nicolaou, et al., *A consensus exercise identifying priorities for research into clinical effectiveness among children's orthopaedic surgeons in the United Kingdom.* Bone Joint J, 2018. **100-b**(5): p. 680-684.

34. Galloway, A.M., T. van-Hille, D.C. Perry, C. Holton, L. Mason, S. Richards, et al., *A systematic review of the non-surgical treatment of Perthes' disease.* 2020. **1**(12): p. 720-730.

35. Biggane, A.M., M. Olsen, and P.R. Williamson, *PPI in research: a reflection from early stage researchers.* Research Involvement and Engagement, 2019. **5**(1): p. 35.

36. Dawson, S., A. Ruddock, V. Parmar, R. Morris, S. Cheraghi-Sohi, S. Giles, et al., *Patient and public involvement in doctoral research: reflections and experiences of the PPI contributors and researcher.* Research Involvement and Engagement, 2020. **6**(1): p. 23.

37. GenerationR. *Young People Improving Research*. 2020 [cited 2024 23/10/2024]; Available from: https://generationr.org.uk/about/.

38. Allemang, B., K. Sitter, and G. Dimitropoulos, *Pragmatism as a paradigm for patient-oriented research.* Health Expect, 2022. **25**(1): p. 38-47.

39. Berryman, D.R., *Ontology, Epistemology, Methodology, and Methods: Information for Librarian Researchers.* Medical Reference Services Quarterly, 2019. **38**(3): p. 271-279.
40. Al-Ababneh, M.M., *Linking ontology, epistemology and research methodology.* Science & Philosophy, 2020. **8**(1): p. 75-91.
41. Creswell, J.W. and V.L. Plano Clark, *Designing and conducting mixed methods research*. Second edition. ed. 2011, Los Angeles, CA ;: SAGE.
42. Shaw, J.A., D.M. Connelly, and A.A. Zecevic, *Pragmatism in practice: mixed methods research for physiotherapy.* Physiother Theory Pract, 2010. **26**(8): p. 510-8.
43. Wilson, C., G. Janes, and J. Williams, *Identity, positionality and reflexivity: relevance and application to research paramedics.* Br Paramed J, 2022. **7**(2): p. 43-49.
44. Skivington, K., L. Matthews, S.A. Simpson, P. Craig, J. Baird, J.M. Blazeby, et al., *A new framework for developing and evaluating complex interventions: update of Medical Research Council guidance.* BMJ, 2021. **374**: p. n2061.
45. Rubin, M. and C. Donkin, *Exploratory hypothesis tests can be more compelling than confirmatory hypothesis tests.* Philosophical Psychology, 2024. **37**(8): p. 2019-2047.
46. Barroga, E. and G.J. Matanguihan, *A Practical Guide to Writing Quantitative and Qualitative Research Questions and Hypotheses in Scholarly Articles.* J Korean Med Sci, 2022. **37**(16): p. e121.
47. Joseph, B. and C.T. Price, *Principles of Containment Treatment Aimed at Preventing Femoral Head Deformation in Perthes Disease.* Orthopedic Clinics of North America, 2011. **42**(3): p. 317-327.
48. Friedlander, J.K. and D.S. Weiner, *Radiographic results of proximal femoral varus osteotomy in Legg-Calvé-Perthes disease.* J Pediatr Orthop, 2000. **20**(5): p. 566-71.
49. Joseph, B., *Management of Perthes' disease.* Indian J Orthop, 2015. **49**(1): p. 10-6.

50. Louahem M'sabah, D., C. Assi, and J. Cottalorda, *Proximal femoral osteotomies in children.* Orthopaedics & Traumatology: Surgery & Research, 2013. **99**(1, Supplement): p. S171-S186.

51. Shah, H., N.D. Siddesh, B. Joseph, and S.N. Nair, *Effect of prophylactic trochanteric epiphyseodesis in older children with Perthes' disease.* J Pediatr Orthop, 2009. **29**(8): p. 889-95.

52. Vogt, B., G. Gosheger, T. Wirth, J. Horn, and R. Rödl, *Leg Length Discrepancy-Treatment Indications and Strategies.* Dtsch Arztebl Int, 2020. **117**(24): p. 405-411.

53. Terjesen, T., O. Wiig, and S. Svenningsen, *Varus femoral osteotomy improves sphericity of the femoral head in older children with severe form of Legg-Calvé-Perthes disease.* Clin Orthop Relat Res, 2012. **470**(9): p. 2394-401.

54. Parmentier, C., A. Madoki, P. Mercier, and P.-L. Docquier, *Shelf acetabuloplasty in Perthes disease: comparison with nonoperative treatment.* Current Orthopaedic Practice, 2016. **27**(4): p. 375-381.

55. Yoo, W.J., I.H. Choi, T.J. Cho, C.Y. Chung, Y.W. Shin, and S.J. Shin, *Shelf acetabuloplasty for children with Perthes' disease and reducible subluxation of the hip: prognostic factors related to hip remodelling.* J Bone Joint Surg Br, 2009. **91**(10): p. 1383-7.

56. Li, W.C. and R.J. Xu, *Lateral shelf acetabuloplasty for severe Legg-Calvé-Perthes disease in patients older than 8 years: A mean eleven-year follow-up.* Medicine (Baltimore), 2016. **95**(45): p. e5272.

57. Ghanem, I., E. Haddad, R. Haidar, S. Haddad-Zebouni, N. Aoun, F. Dagher, et al., *Lateral shelf acetabuloplasty in the treatment of Legg-Calvé-Perthes disease: improving mid-term outcome in severely deformed hips.* J Child Orthop, 2010. **4**(1): p. 13-20.

58. Freeman, R.T., A.M. Wainwright, T.N. Theologis, and M.K. Benson, *The Outcome of Patients With Hinge Abduction in Severe Perthes Disease Treated by Shelf Acetabuloplasty.* Journal of Pediatric Orthopaedics, 2008. **28**(6): p. 619-625.

59. Segev, E., E. Ezra, S. Wientroub, M. Yaniv, S. Hayek, and Y. Hemo, *Treatment of severe late-onset Perthes' disease with soft tissue release and articulated hip distraction: revisited at skeletal maturity.* J Child Orthop, 2007. **1**(4): p. 229-35.

60. Zhamilov, V., C.D. Basa, İ.E. Kaçmaz, A. Reisoğlu, and H. Agus, *Functional Outcomes of Soft Tissue Release Surgery in Advanced Legg-Calve-Perthes Disease.* Turk J Hip Surg, 2021. **1**(3): p. 84-89.

61. Brech, G.C. and R. Guarnieiro, *Evaluation of physiotherapy in the treatment of Legg-Calvé-Perthes disease.* Clinics (Sao Paulo), 2006. **61**(6): p. 521-8.

62. Wells, G.A., B. Shea, D. O'Connell, J. Peterson, V. Welch, M. Losos, et al., *The Newcastle-Ottawa Scale (NOS) for assessing the quality of nonrandomised studies in meta-analyses.* 2000.

63. Hoffmann, T.C., P.P. Glasziou, I. Boutron, R. Milne, R. Perera, D. Moher, et al., *Better reporting of interventions: template for intervention description and replication (TIDieR) checklist and guide.* Bmj, 2014. **348**: p. g1687.

64. Rich, M.M. and P.L. Schoenecker, *Management of Legg-Calvé-Perthes Disease Using an A-Frame Orthosis and Hip Range of Motion: A 25-Year Experience.* Journal of pediatric orthopaedics, 2013. **33**(2): p. 112-119.

65. Gobeil-Lavoie, A.-P., M.-C. Chouinard, A. Danish, and C. Hudon, *Characteristics of self-management among patients with complex health needs: a thematic analysis review.* BMJ Open, 2019. **9**(5): p. e028344.

66. Lorig, K.R. and H.R. Holman, *Self-management education: History, definition, outcomes, and mechanisms.* Annals of Behavioral Medicine, 2003. **26**(1): p. 1-7.

67. England, N. *Supported self-management*. 2020 [cited 2024 15/10/2024]; Available from: https://www.england.nhs.uk/personalisedcare/supported-self-management/.

68. Modi, A.C., A.L. Pai, K.A. Hommel, K.K. Hood, S. Cortina, M.E. Hilliard, et al., *Pediatric Self-management: A Framework for Research, Practice, and Policy.* Pediatrics, 2012. **129**(2): p. e473-e485.

69. Du, S., C. Yuan, X. Xiao, J. Chu, Y. Qiu, and H. Qian, *Self-management programs for chronic musculoskeletal pain conditions: a systematic review and meta-analysis.* Patient Educ Couns, 2011. **85**(3): p. e299-310.

70. Effing, T., E.E. Monninkhof, P.P. Van Der Valk, G.G. Zielhuis, E.H. Walters, J.J. Van Der Palen, et al., *Self-management education for patients with chronic obstructive pulmonary disease.* Cochrane Database of Systematic Reviews, 2007(4).

71. Jovicic, A., J.M. Holroyd-Leduc, and S.E. Straus, *Effects of self-management intervention on health outcomes of patients with heart failure: a systematic review of randomized controlled trials.* BMC cardiovascular disorders, 2006. **6**: p. 1-8.

72. Swindle, T., A.B. Poosala, N. Zeng, E. Børsheim, A. Andres, and L.L. Bellows, *Digital Intervention Strategies for Increasing Physical Activity Among Preschoolers: Systematic Review.* J Med Internet Res, 2022. **24**(1): p. e28230.

73. Mannocci, A., V. D'Egidio, I. Backhaus, A. Federici, A. Sinopoli, A. Ramirez Varela, et al., *Are There Effective Interventions to Increase Physical Activity in Children and Young People? An Umbrella Review.* Int J Environ Res Public Health, 2020. **17**(10).

74. Bleich, S.N., K.A. Vercammen, L.Y. Zatz, J.M. Frelier, C.B. Ebbeling, and A. Peeters, *Interventions to prevent global childhood overweight and obesity: a systematic review.* Lancet Diabetes Endocrinol, 2018. **6**(4): p. 332-346.

75. Brown, H.E., A.J. Atkin, J. Panter, G. Wong, M.J. Chinapaw, and E.M. van Sluijs, *Family-based interventions to increase physical activity in children: a systematic review, meta-analysis and realist synthesis.* Obes Rev, 2016. **17**(4): p. 345-60.

76. Dellert, J.C. and P. Johnson, *Interventions with Children and Parents to Improve Physical Activity and Body Mass Index: A Meta-Analysis.* American Journal of Health Promotion, 2014. **28**(4): p. 259-267.

77. Metcalf, B., W. Henley, and T. Wilkin, *Effectiveness of intervention on physical activity of children: systematic review and meta-analysis of controlled trials*

with objectively measured outcomes (EarlyBird 54). BMJ : British Medical Journal, 2012. **345**: p. e5888.

78. Teixeira, P.J., E.V. Carraça, D. Markland, M.N. Silva, and R.M. Ryan, *Exercise, physical activity, and self-determination theory: A systematic review.* International Journal of Behavioral Nutrition and Physical Activity, 2012. **9**(1): p. 78.
79. Hamel, L.M. and L.B. Robbins, *Computer- and web-based interventions to promote healthy eating among children and adolescents: a systematic review.* J Adv Nurs, 2013. **69**(1): p. 16-30.
80. Lau, P.W., E.Y. Lau, P. Wong del, and L. Ransdell, *A systematic review of information and communication technology-based interventions for promoting physical activity behavior change in children and adolescents.* J Med Internet Res, 2011. **13**(3): p. e48.
81. McIntosh, J.R.D., S. Jay, N. Hadden, and P.J. Whittaker, *Do E-health interventions improve physical activity in young people: a systematic review.* Public Health, 2017. **148**: p. 140-148.
82. Monroe, C.M., D.L. Thompson, D.R. Bassett, E.C. Fitzhugh, and H.A. Raynor, *Usability of Mobile Phones in Physical Activity–Related Research: A Systematic Review.* American Journal of Health Education, 2015. **46**(4): p. 196-206.
83. Pakarinen, A., H. Parisod, J. Smed, and S. Salanterä, *Health game interventions to enhance physical activity self-efficacy of children: a quantitative systematic review.* J Adv Nurs, 2017. **73**(4): p. 794-811.
84. Schoeppe, S., S. Alley, W. Van Lippevelde, N.A. Bray, S.L. Williams, M.J. Duncan, et al., *Efficacy of interventions that use apps to improve diet, physical activity and sedentary behaviour: a systematic review.* Int J Behav Nutr Phys Act, 2016. **13**(1): p. 127.
85. Shea, B.J., J.M. Grimshaw, G.A. Wells, M. Boers, N. Andersson, C. Hamel, et al., *Development of AMSTAR: a measurement tool to assess the methodological quality of systematic reviews.* BMC Med Res Methodol, 2007. **7**: p. 10.

86. Shea, B.J., C. Hamel, G.A. Wells, L.M. Bouter, E. Kristjansson, J. Grimshaw, et al., *AMSTAR is a reliable and valid measurement tool to assess the methodological quality of systematic reviews.* J Clin Epidemiol, 2009. **62**(10): p. 1013-20.

87. Shea, B.J., B.C. Reeves, G. Wells, M. Thuku, C. Hamel, J. Moran, et al., *AMSTAR 2: a critical appraisal tool for systematic reviews that include randomised or non-randomised studies of healthcare interventions, or both.* BMJ, 2017. **358**: p. j4008.

88. (NICE), N.I.f.H.a.C.E. *Evidence standards framework for Digital Health Technologies*. 2019 [cited 2020 27/05/2020]; Available from: https://www.nice.org.uk/Media/Default/About/what-we-do/our-programmes/evidence-standards-framework/digital-evidence-standards-framework.pdf.

89. (NICE), N.I.f.H.a.C.E. *Evidence standards framework for Digital Health Technologies*. 2022 [cited 2024 03/09/2024]; Available from: https://www.nice.org.uk/corporate/ecd7/resources/evidence-standards-framework-for-digital-health-technologies-pdf-1124017457605.

90. Webbe, J., I. Sinha, and C. Gale, *Core Outcome Sets.* Archives of disease in childhood - Education & practice edition, 2018. **103**(3): p. 163-166.

91. Williamson, P.R., D.G. Altman, H. Bagley, K.L. Barnes, J.M. Blazeby, S.T. Brookes, et al., *The COMET Handbook: version 1.0.* Trials, 2017. **18**(3): p. 280.

92. Hughes, K.L., J.J. Kirkham, M. Clarke, and P.R. Williamson, *Assessing the impact of a research funder's recommendation to consider core outcome sets.* PLoS One, 2019. **14**(9): p. e0222418.

93. Research, N.I.f.H.a.C. *HTA Programme supporting information (REALMS)*. 2022 [cited 2024 03/09/2024]; Available from: https://www.nihr.ac.uk/documents/hta-supporting-information/27265.

94. Ollivere, B.J., B.A. Marson, and F.S. Haddad, *Getting the right answer.* The Bone & Joint Journal, 2019. **101-B**(3): p. 233-235.

95. Leo, D.G., H. Jones, R. Murphy, J.W. Leong, T. Gambling, A.F. Long, et al., *The outcomes of Perthes' disease.* Bone Joint J, 2020. **102-b**(5): p. 611-617.

96. Boers, M., J.R. Kirwan, G. Wells, D. Beaton, L. Gossec, M.A. d'Agostino, et al., *Developing core outcome measurement sets for clinical trials: OMERACT filter 2.0.* J Clin Epidemiol, 2014. **67**(7): p. 745-53.

97. Calvert, M., D. Kyte, R. Mercieca-Bebber, A. Slade, A.-W. Chan, M.T. King, et al., *Guidelines for Inclusion of Patient-Reported Outcomes in Clinical Trial Protocols: The SPIRIT-PRO Extension.* JAMA, 2018. **319**(5): p. 483-494.

98. Broderick, J.E., E.M. DeWitt, N. Rothrock, P.K. Crane, and C.B. Forrest, *Advances in Patient-Reported Outcomes: The NIH PROMIS(®) Measures.* EGEMS (Wash DC), 2013. **1**(1): p. 1015.

99. Brandon, T.G., B.D. Becker, K.B. Bevans, and P.F. Weiss, *Patient-Reported Outcomes Measurement Information System Tools for Collecting Patient-Reported Outcomes in Children With Juvenile Arthritis.* Arthritis Care Res (Hoboken), 2017. **69**(3): p. 393-402.

100. Howell, C.R., L.A. Thompson, H.E. Gross, B.B. Reeve, D.A. DeWalt, and I.C. Huang, *Responsiveness to Change in PROMIS(®) Measures among Children with Asthma: A Report from the PROMIS(®) Pediatric Asthma Study.* Value Health, 2016. **19**(2): p. 192-201.

101. Nguyen, T.H., H.R. Han, M.T. Kim, and K.S. Chan, *An introduction to item response theory for patient-reported outcome measurement.* Patient, 2014. **7**(1): p. 23-35.

102. Matsumoto, H., J.E. Hyman, H.H. Shah, W.N. Sankar, J.C. Laine, C.T. Mehlman, et al., *Validation of Pediatric Self-Report Patient-Reported Outcomes Measurement Information System (PROMIS) Measures in Different Stages of Legg-Calvé-Perthes Disease.* J Pediatr Orthop, 2020. **40**(5): p. 235-240.

103. Luo, W., M.S. Ali, R. Limb, C. Cornforth, and D.C. Perry, *Use of the PROMIS Mobility score in assessing function in adolescents and adults previously affected by childhood hip disease.* Bone Jt Open, 2021. **2**(12): p. 1089-1095.

104. Moore, M.E., D.M. Vega, K.M. Wiens, and N. Caporale, *Connecting Theory to Practice: Using Self-Determination Theory To Better Understand Inclusion in STEM.* Journal of microbiology & biology education, 2020. **21**(1): p. 21.1.32.

105. Klos, L., K. Feil, T. Eberhardt, and D. Jekauc, *Interventions to Promote Positive Affect and Physical Activity in Children, Adolescents and Young Adults-A Systematic Review.* Sports (Basel, Switzerland), 2020. **8**(2): p. 26.

106. Cox, A., N. Duncheon, and L. McDavid, *Peers and teachers as sources of relatedness perceptions, motivation, and affective responses in physical education.* Res Q Exerc Sport, 2009. **80**(4): p. 765-73.

107. Gråstén, A. and A. Watt, *A Motivational Model of Physical Education and Links to Enjoyment, Knowledge, Performance, Total Physical Activity and Body Mass Index.* J Sports Sci Med, 2017. **16**(3): p. 318-327.

108. Murillo Pardo, B., E. García Bengoechea, J.A. Julián Clemente, and E. Generelo Lanaspa, *Motivational Outcomes and Predictors of Moderate-to-Vigorous Physical Activity and Sedentary Time for Adolescents in the Sigue La Huella Intervention.* Int J Behav Med, 2016. **23**(2): p. 135-42.

109. Mehtälä, M.A.K., A.K. Sääkslahti, M.E. Inkinen, and M.E.H. Poskiparta, *A socio-ecological approach to physical activity interventions in childcare: a systematic review.* International Journal of Behavioral Nutrition and Physical Activity, 2014. **11**(1): p. 22.

110. Zhang, T. and M. Solmon, *Integrating self-determination theory with the social ecological model to understand students' physical activity behaviors.* International Review of Sport and Exercise Psychology, 2013. **6**(1): p. 54-76.

111. Gale, N.K., G. Heath, E. Cameron, S. Rashid, and S. Redwood, *Using the framework method for the analysis of qualitative data in multi-disciplinary health research.* BMC Medical Research Methodology, 2013. **13**(1): p. 117.

112. Morison, M., J. Moir, and T. Kwansa, *Interviewing children for the purposes of research in primary care.* Primary Health Care Research & Development, 2000. **1**(2): p. 113-130.

113. Leung, F.H. and R. Savithiri, *Spotlight on focus groups.* Can Fam Physician, 2009. **55**(2): p. 218-9.

114. O.Nyumba, T., K. Wilson, C.J. Derrick, and N. Mukherjee, *The use of focus group discussion methodology: Insights from two decades of application in conservation.* Methods in Ecology and Evolution, 2018. **9**(1): p. 20-32.

115. Pini, S., P. Gardner, and S. Hugh-Jones, *How and Why School Is Important to Teenagers with Cancer: Outcomes from a Photo-Elicitation Study.* J Adolesc Young Adult Oncol, 2019. **8**(2): p. 157-164.

116. Pini, S., S. Hugh-Jones, L. Shearsmith, and P. Gardner, *'What are you crying for? I don't even know you' – The experiences of teenagers communicating with their peers when returning to school.* European Journal of Oncology Nursing, 2019. **39**: p. 28-34.

117. Ponizovsky-Bergelson, Y., Y. Dayan, N. Wahle, and D. Roer-Strier, *A Qualitative Interview With Young Children: What Encourages or Inhibits Young Children's Participation?* International Journal of Qualitative Methods, 2019. **18**: p. 1609406919840516.

118. West, C.H., D.L. Dusome, J. Winsor, A. Winther Klippenstein, and L.B. Rallison, *Dialoguing With Images: An Expressive Arts Method for Health Research.* Qual Health Res, 2022. **32**(7): p. 1055-1070.

119. Ritchie, J., J. Lewis, C.M. Nicholls, and R. Ormston. *Qualitative Research Practice: A Guide for Social Science Students and Researchers*. 2013.

120. Furber, C., *Framework analysis: a method for analysing qualitative data.* African Journal of Midwifery and Women's Health, 2010. **4**(2): p. 97-100.

121. Smith, J. and J. Firth, *Qualitative data analysis: the framework approach.* Nurse Res, 2011. **18**(2): p. 52-62.

122. Fereday, J. and E. Muir-Cochrane, *Demonstrating Rigor Using Thematic Analysis: A Hybrid Approach of Inductive and Deductive Coding and Theme Development.* International Journal of Qualitative Methods, 2006. **5**(1): p. 80-92.

123. Elo, S., M. Kääriäinen, O. Kanste, T. Pölkki, K. Utriainen, and H. Kyngäs, *Qualitative Content Analysis: A Focus on Trustworthiness.* SAGE Open, 2014. **4**(1): p. 2158244014522633.

124. Mayring, P., *Qualitative content analysis: Theoretical background and procedures*, in *Approaches to qualitative research in mathematics education*. 2015, Springer. p. 365-380.

125. Braun, V. and V. Clarke, *Using thematic analysis in psychology.* Qualitative Research in Psychology, 2006. **3**(2): p. 77-101.

126. Vaismoradi, M., H. Turunen, and T. Bondas, *Content analysis and thematic analysis: Implications for conducting a qualitative descriptive study.* Nursing & Health Sciences, 2013. **15**(3): p. 398-405.

127. Murphy, M.K., N.A. Black, D.L. Lamping, C.M. McKee, C.F. Sanderson, J. Askham, et al., *Consensus development methods, and their use in clinical guideline development.* Health Technol Assess, 1998. **2**(3): p. i-iv, 1-88.

128. Goodman, C.S. and S.R. Baratz. *Improving Consensus Development for Health Technology Assessment: An International Perspective*. 1990.

129. Manera, K., C.S. Hanson, T. Gutman, and A. Tong, *Consensus Methods: Nominal Group Technique*, in *Handbook of Research Methods in Health Social Sciences*, P. Liamputtong, Editor. 2019, Springer Singapore: Singapore. p. 737-750.

130. Mallett, R., S. McLean, M.A. Holden, T. Potia, M. Gee, and K. Haywood, *Use of the nominal group technique to identify UK stakeholder views of the measures and domains used in the assessment of therapeutic exercise adherence for patients with musculoskeletal disorders.* BMJ Open, 2020. **10**(2): p. e031591.

131. Arakawa, N. and L.R. Bader, *Consensus development methods: Considerations for national and global frameworks and policy development.* Research in Social and Administrative Pharmacy, 2022. **18**(1): p. 2222-2229.

132. Murray, I.R., N.S. Makaram, R.F. LaPrade, and F.S. Haddad, *Consensus statements: when and how?* Bone Joint J, 2023. **105-b**(4): p. 343-346.

133. Sparks, J.B., M.L. Klamerus, T.J. Caverly, S.E. Skurla, T.P. Hofer, E.A. Kerr, et al., *Planning and Reporting Effective Web-Based RAND/UCLA Appropriateness Method Panels: Literature Review and Preliminary Recommendations.* J Med Internet Res, 2022. **24**(8): p. e33898.

134. Eubank, B.H., N.G. Mohtadi, M.R. Lafave, J.P. Wiley, A.J. Bois, R.S. Boorman, et al., *Using the modified Delphi method to establish clinical consensus for the diagnosis and treatment of patients with rotator cuff pathology.* BMC Medical Research Methodology, 2016. **16**(1): p. 56.

135. Magnusson, D.M., Z.D. Rethorn, E.H. Bradford, J. Maxwell, M.S. Ingman, T.E. Davenport, et al., *Population Health, Prevention, Health Promotion, and Wellness Competencies in Physical Therapist Professional Education: Results of a Modified Delphi Study.* Physical Therapy, 2020. **100**(9): p. 1645-1658.

136. Nasa, P., R. Jain, and D. Juneja, *Delphi methodology in healthcare research: How to decide its appropriateness.* World journal of methodology, 2021. **11**(4): p. 116-129.

137. Wiangkham, T., J. Duda, M.S. Haque, and A. Rushton, *Development of an active behavioural physiotherapy intervention (ABPI) for acute whiplash-associated disorder (WAD) II management: a modified Delphi study.* BMJ Open, 2016. **6**(9): p. e011764.

138. O'Cathain, A., L. Croot, K. Sworn, E. Duncan, N. Rousseau, K. Turner, et al., *Taxonomy of approaches to developing interventions to improve health: a systematic methods overview.* Pilot and Feasibility Studies, 2019. **5**(1): p. 41.

139. Gentles, S.J., C. Charles, D.B. Nicholas, J. Ploeg, and K.A. McKibbon, *Reviewing the research methods literature: principles and strategies illustrated by a systematic overview of sampling in qualitative research.* Systematic Reviews, 2016. **5**(1): p. 172.

140. Mummah, S.A., T.N. Robinson, A.C. King, C.D. Gardner, and S. Sutton, *IDEAS (Integrate, Design, Assess, and Share): A Framework and Toolkit of Strategies for the Development of More Effective Digital Interventions to Change Health Behavior.* J Med Internet Res, 2016. **18**(12): p. e317.

141. Reed, H., D. Couturiaux, M. Davis, A. Edwards, E. Janes, H.S. Kim, et al., *Co-production as an Emerging Methodology for Developing School-Based Health Interventions with Students Aged 11-16: Systematic Review of Intervention Types, Theories and Processes and Thematic Synthesis of Stakeholders' Experiences.* Prev Sci, 2021. **22**(4): p. 475-491.

142. Reed, H., S. Murphy, and R. Evans, *Optimising a co-production framework for developing public health interventions: application and testing of school-based Research Action Groups.* Health Research Policy and Systems, 2023. **21**(1): p. 133.

143. Santina, T., D. Beaulieu, C. Gagné, and L. Guillaumie, *Using the intervention mapping protocol to promote school-based physical activity among children: A demonstration of the step-by-step process.* Health Education Journal, 2020. **79**(5): p. 529-542.

144. Jones, T.M., B.F. Dear, J.M. Hush, N. Titov, and C.M. Dean, *Application of Intervention Mapping to the Development of a Complex Physical Therapist Intervention.* Physical Therapy, 2016. **96**(12): p. 1994-2004.

145. Michie, S., M.M. van Stralen, and R. West, *The behaviour change wheel: A new method for characterising and designing behaviour change interventions.* Implementation Science, 2011. **6**(1): p. 42.

146. O'Cathain, A., E. Murphy, and J. Nicholl, *Why, and how, mixed methods research is undertaken in health services research in England: a mixed methods study.* BMC Health Services Research, 2007. **7**(1): p. 85.

147. Ivankova, N.V., J.W. Creswell, and S.L. Stick, *Using Mixed-Methods Sequential Explanatory Design: From Theory to Practice.* Field Methods, 2006. **18**(1): p. 3-20.

148. study, T.C. *The CRAFFT Study*. 2024 [cited 2024 03/07/2024]; Available from: https://crafft-study.digitrial.com.

149. Phelps, E.E., E. Tutton, M.L. Costa, J. Achten, P. Gibson, A. Moscrop, et al., *Being recovered: a qualitative study of parents' experience of their child's recovery up to a year after a displaced distal radius fracture.* Bone & Joint Open, 2024. **5**(5): p. 426-434.

150. Kettles, A.M., J.W. Cresswell, and W. Zhang, *Mixed methods research in mental health nursing.* Journal of Psychiatric and Mental Health Nursing, 2011. **18**(6): p. 535-542.

151. Vedel, I., N. Kaur, Q.N. Hong, R. El Sherif, V. Khanassov, C. Godard-Sebillotte, et al., *Why and how to use mixed methods in primary health care research.* Fam Pract, 2019. **36**(3): p. 365-368.

152. Eldridge, S.M., G.A. Lancaster, M.J. Campbell, L. Thabane, S. Hopewell, C.L. Coleman, et al., *Defining Feasibility and Pilot Studies in Preparation for*

Randomised Controlled Trials: Development of a Conceptual Framework. PLoS One, 2016. **11**(3): p. e0150205.

153. Gill, P., K. Stewart, E. Treasure, and B. Chadwick, *Methods of data collection in qualitative research: interviews and focus groups.* British Dental Journal, 2008. **204**(6): p. 291-295.

154. Kennedy, C., S. Kools, and R. Krueger, *Methodological considerations in children's focus groups.* Nurs Res, 2001. **50**(3): p. 184-7.

155. Galloway, A.M., S. Pini, C. Holton, D.C. Perry, A. Redmond, H.J. Siddle, et al., *"Waiting for the best day of your life". A qualitative interview study of patients' and clinicians' experiences of Perthes' disease.* Bone & Joint Open, 2023. **4**(10): p. 735-741.

156. Brech, G.C. and R. Guarnieiro, *Evaluation of physiotherapy in the treatment of Legg-Calve-Perthes disease.* Clinics, 2006. **61**(6): p. 521-528.

157. Hills, A.P., L.B. Andersen, and N.M. Byrne, *Physical activity and obesity in children.* 2011. **45**(11): p. 866-870.

158. Morton, K., L. Dennison, C. May, E. Murray, P. Little, R.J. McManus, et al., *Using digital interventions for self-management of chronic physical health conditions: A meta-ethnography review of published studies.* Patient Education and Counseling, 2017. **100**(4): p. 616-635.

159. Leo, D.G., R. Murphy, T. Gambling, A. Long, H. Jones, and D.C. Perry, *Perspectives on the Social, Physical, and Emotional Impact of Living With Perthes' Disease in Children and Their Family: A Mixed Methods Study.* Global Pediatric Health, 2019. **6**: p. 2333794X19835235.

160. Leo, D.G., H. Jones, R. Murphy, J.W. Leong, T. Gambling, A.F. Long, et al., *The outcomes of Perthes' disease.* The Bone & Joint Journal, 2020. **102-B**(5): p. 611-617.

161. Almoajil, H., T. Theologis, H. Dawes, J. Pierce, A. Meaney, A. Baklouti, et al., *Exploring the factors that influence stakeholders' expectations and subsequent perception of lower limb orthopaedic surgical outcomes for ambulant children with cerebral palsy – a qualitative study.* Disability and Rehabilitation, 2022: p. 1-8.

162. Papiez, K., E. Tutton, E.E. Phelps, J. Baird, M.L. Costa, J. Achten, et al., *A qualitative study of parents' and their child's experience of a medial epicondyle fracture.* Bone Jt Open, 2021. **2**(6): p. 359-364.

163. Warburton, D.E.R. and S.S.D. Bredin, *Health Benefits of Physical Activity: A Strengths-Based Approach.* Journal of clinical medicine, 2019. **8**(12): p. 2044.

164. Yardley, L., B. Ainsworth, E. Arden-Close, and I. Muller, *The person-based approach to enhancing the acceptability and feasibility of interventions.* Pilot and Feasibility Studies, 2015. **1**(1): p. 37.

165. Yardley, L., L. Morrison, K. Bradbury, and I. Muller, *The person-based approach to intervention development: application to digital health-related behavior change interventions.* Journal of medical Internet research, 2015. **17**(1): p. e30-e30.

166. Kolehmainen, N., J.J. Francis, C.R. Ramsay, C. Owen, L. McKee, M. Ketelaar, et al., *Participation in physical play and leisure: developing a theory- and evidence-based intervention for children with motor impairments.* BMC Pediatr, 2011. **11**: p. 100.

167. Kolehmainen, N., C. Ramsay, L. McKee, C. Missiuna, C. Owen, and J. Francis, *Participation in Physical Play and Leisure in Children With Motor Impairments: Mixed-Methods Study to Generate Evidence for Developing an Intervention.* Phys Ther, 2015. **95**(10): p. 1374-86.

168. Teherani, A., T. Martimianakis, T. Stenfors-Hayes, A. Wadhwa, and L. Varpio, *Choosing a Qualitative Research Approach.* J Grad Med Educ, 2015. **7**(4): p. 669-70.

169. Chun Tie, Y., M. Birks, and K. Francis, *Grounded theory research: A design framework for novice researchers.* SAGE Open Med, 2019. **7**: p. 2050312118822927.

170. Strudwick, R.M., *Ethnographic research in healthcare - patients and service users as participants.* Disabil Rehabil, 2021. **43**(22): p. 3271-3275.

171. Blackstone, A., *Principles of sociological inquiry: Qualitative and quantitative methods*. 2018: Saylor Academy Open Textbooks.

172. Azungah, T. and R. Kasmad, *Qualitative Research Journal Article information: For Authors Qualitative research: deductive and inductive approaches to data analysis.* 2020.

173. Coyne, I.T., *Sampling in qualitative research. Purposeful and theoretical sampling; merging or clear boundaries?* 1997. **26**(3): p. 623-630.

174. Authority, N.H.R. *Applying a proportionate approach to the process of seeking consent*. 2017 [cited 2021 09/06/2021]; Available from: https://www.hra.nhs.uk/documents/283/applying-proportionate-approach-process-seeking-consent_R3gbJKn.pdf.

175. NVivo. *NVivo software*. 2020 [cited 2023 26/01/2023]; Available from: https://www.qsrinternational.com/nvivo-qualitative-data-analysis-software/enabling-research/nvivo-getting-started-bundle.

176. NHS. *NHS Health Research Authority - Student research*. 2021 [cited 2021 21/05/2021]; Available from: https://www.hra.nhs.uk/planning-and-improving-research/research-planning/student-research/.

177. Authority, N.H.R. *Research involving children* 2021 06/09/2021 [cited 2022 22/02/2022]; Available from: https://www.hra.nhs.uk/planning-and-improving-research/policies-standards-legislation/research-involving-children/.

178. Manti, S. and A. Licari, *How to obtain informed consent for research.* Breathe (Sheff), 2018. **14**(2): p. 145-152.

179. Baines, P., *Assent for children's participation in research is incoherent and wrong.* Archives of Disease in Childhood, 2011. **96**(10): p. 960-962.

180. Waligora, M., V. Dranseika, and J. Piasecki, *Child's assent in research: Age threshold or personalisation?* BMC Medical Ethics, 2014. **15**(1): p. 44.

181. Alliance, J.L. *Priority 4 from the Paediatric Lower Limb Surgery PSP*. 2018 [cited 2020 03/06/2020]; Available from: http://www.jla.nihr.ac.uk/priority-setting-partnerships/Paediatric-lower-limb-surgery/priority-4-from-the-paediatric-lower-limb-surgery-psp.htme.

182. NHS. *Chapter 5: Digitally enabled care will go mainstream across the NHS*. 2018 [cited 2020 03/06/2020]; Available from:

https://www.longtermplan.nhs.uk/online-version/chapter-5-digitally-enabled-care-will-go-mainstream-across-the-nhs/.

183. Finlay, L., *Reflexivity: An Essential Component for All Research?* British Journal of Occupational Therapy, 1998. **61**(10): p. 453-456.

184. Macbeth, D., *On "Reflexivity" in Qualitative Research: Two Readings, and a Third.* Qualitative Inquiry, 2001. **7**(1): p. 35-68.

185. Waggoner, J., J.D. Carline, and S.J. Durning, *Is There a Consensus on Consensus Methodology? Descriptions and Recommendations for Future Consensus Research.* Acad Med, 2016. **91**(5): p. 663-8.

186. Akins, R.B., H. Tolson, and B.R. Cole, *Stability of response characteristics of a Delphi panel: application of bootstrap data expansion.* BMC Med Res Methodol, 2005. **5**: p. 37.

187. Gelfer, Y., J. Blanco, A. Trees, N. Davis, R. Buckingham, A.C. Peek, et al., *Attaining a British consensus statement on managing idiopathic congenital talipes equinovarus (CTEV) through a Delphi process: a study protocol.* BMJ Open, 2021. **11**(9): p. e049212.

188. Vogel, C., S. Zwolinsky, C. Griffiths, M. Hobbs, E. Henderson, and E. Wilkins, *A Delphi study to build consensus on the definition and use of big data in obesity research.* International Journal of Obesity, 2019. **43**(12): p. 2573-2586.

189. Mitchell, P.D., A. Abraham, C. Carpenter, P.D. Henman, J. Mavrotas, J. McCaul, et al., *Consensus guidelines on the management of musculoskeletal infection affecting children in the UK.* Bone Joint J, 2023. **105-b**(7): p. 815-820.

190. Surgery, B.S.f.C.s.O. *BSCOS DDH Consensus Group* 2022 [cited 2023 18/04/2023]; Available from: https://www.bscos.org.uk/consensus/consensus/DDH.php.

191. Davids, J.R., J. Shilt, R. Kay, T. Dreher, B.J. Shore, J. McCarthy, et al., *Assessment of foot alignment and function for ambulatory children with cerebral palsy: Results of a modified Delphi technique consensus study.* Journal of Children's Orthopaedics, 2022. **16**(2): p. 111-120.

192. Rodríguez-Olivas, A.O., E. Hernández-Zamora, and E. Reyes-Maldonado, *Legg-Calvé-Perthes disease overview.* Orphanet J Rare Dis, 2022. **17**(1): p. 125.

193. Jisc. *Online surveys: Powerful, flexible online surveys*. 2022 [cited 2022 13/05/2022]; Available from: https://www.onlinesurveys.ac.uk.

194. Beiderbeck, D., N. Frevel, H.A. von der Gracht, S.L. Schmidt, and V.M. Schweitzer, *Preparing, conducting, and analyzing Delphi surveys: Cross-disciplinary practices, new directions, and advancements.* MethodsX, 2021. **8**: p. 101401.

195. Nair, R., R. Aggarwal, and D. Khanna, *Methods of formal consensus in classification/diagnostic criteria and guideline development.* Semin Arthritis Rheum, 2011. **41**(2): p. 95-105.

196. Landis, J.R. and G.G. Koch, *The measurement of observer agreement for categorical data.* Biometrics, 1977. **33**(1): p. 159-74.

197. Dagenais, F., *The Reliability and Convergence of the Delphi Technique.* The Journal of General Psychology, 1978. **98**(2): p. 307-308.

198. Hsu, C.-C. and B.A. Sandford, *The Delphi technique: making sense of consensus.* Practical assessment, research, and evaluation, 2007. **12**(1): p. 10.

199. Linstone, H.A. and M. Turoff, *The Delphi Method: Techniques and Applications*. 2002: Addison-Wesley Publishing Company, Advanced Book Program.

200. Anderson, A.M., C. Comer, T.O. Smith, B.T. Drew, H. Pandit, D. Antcliff, et al., *Consensus on pre-operative total knee replacement education and prehabilitation recommendations: a UK-based modified Delphi study.* BMC Musculoskeletal Disorders, 2021. **22**(1): p. 352.

201. Diamond, I.R., R.C. Grant, B.M. Feldman, P.B. Pencharz, S.C. Ling, A.M. Moore, et al., *Defining consensus: A systematic review recommends methodologic criteria for reporting of Delphi studies.* Journal of Clinical Epidemiology, 2014. **67**(4): p. 401-409.

202. Comer, C., C. Ammendolia, M.C. Battié, A. Bussières, J. Fairbank, A. Haig, et al., *Consensus on a standardised treatment pathway algorithm for lumbar spinal stenosis: an international Delphi study.* BMC Musculoskeletal Disorders, 2022. **23**(1): p. 550.

203. Keeney, S., H.A. McKenna, and F. Hasson, *The Delphi Technique in Nursing and Health Research*. 2011: Wiley.

204. Leeds, U.o. *Guidance for completing the application for for University ethical review*. 2022 January 2016 [cited 2022 13/05/2022]; Available from: https://ris.leeds.ac.uk/wp-content/uploads/2020/07/Guidance_on_Ethical_review_form_V3.pdf.

205. El-Harbiti, A. and Y.D. Hailer, *Range of abduction in patients with Legg-Calvé-Perthes disease – a nationwide register-based cohort study.* BMC Musculoskeletal Disorders, 2020. **21**(1): p. 718.

206. Frasson, V.B., M.A. Vaz, A.B. Morales, A. Torresan, M.A. Telöken, P.D.F. Gusmão, et al., *Hip muscle weakness and reduced joint range of motion in patients with femoroacetabular impingement syndrome: a case-control study.* Braz J Phys Ther, 2020. **24**(1): p. 39-45.

207. Clohisy, J.C., J.R. Ross, J.D. North, J.J. Nepple, and P.L. Schoenecker, *What Are the Factors Associated With Acetabular Correction in Perthes-like Hip Deformities?* Clinical Orthopaedics and Related Research®, 2012. **470**(12): p. 3439-3445.

208. Mazloumi, S.M., M.H. Ebrahimzadeh, and A.R. Kachooei, *Evolution in diagnosis and treatment of Legg-Calve-Perthes disease.* Arch Bone Jt Surg, 2014. **2**(2): p. 86-92.

209. Mills, W., N. Kondakis, R. Orr, M. Warburton, and N. Milne, *Does Hydrotherapy Impact Behaviours Related to Mental Health and Well-Being for Children with Autism Spectrum Disorder? A Randomised Crossover-Controlled Pilot Trial.* International Journal of Environmental Research and Public Health, 2020. **17**(2): p. 558.

210. Epps, H., L. Ginnelly, M. Utley, T. Southwood, S. Gallivan, M. Sculpher, et al., *Is hydrotherapy cost-effective? A randomised controlled trial of combined hydrotherapy programmes compared with physiotherapy land techniques in children with juvenile idiopathic arthritis.* Health technology assessment (Winchester, England), 2005. **9**(39): p. iii-iv, ix-x, 1-59.

211. Kutzner, I., A. Richter, K. Gordt, J. Dymke, P. Damm, G.N. Duda, et al., *Does aquatic exercise reduce hip and knee joint loading? In vivo load measurements with instrumented implants.* PLoS One, 2017. **12**(3): p. e0171972.

212. Kuroda, T., S. Mitani, Y. Sugimoto, K. Asaumi, H. Endo, H. Akazawa, et al., *Changes in the lateral pillar classification in Perthes' disease.* Journal of Pediatric Orthopaedics B, 2009. **18**(3): p. 116-119.

213. Hines, D., T. Ford, S. Westwood, J.R. Barrett, B. Westphal, V. Davies, et al., *Evaluating the provision of paediatric liaison psychiatry services in England.* BJPsych Open, 2023. **9**(2): p. e30.

214. Stanitski, C.L., *Hip range of motion in Perthes' disease: comparison of pre-operative and intra-operative values.* J Child Orthop, 2007. **1**(1): p. 33-5.

215. Tannast, M., M. Hanke, T.M. Ecker, S.B. Murphy, C.E. Albers, and M. Puls, *LCPD: reduced range of motion resulting from extra- and intraarticular impingement.* Clin Orthop Relat Res, 2012. **470**(9): p. 2431-40.

216. Maleki, A., S.M. Qoreishy, and M.N. Bahrami, *Surgical Treatments for Legg-Calvé-Perthes Disease: Comprehensive Review.* Interact J Med Res, 2021. **10**(2): p. e27075.

217. Kleynen, M., S.M. Braun, M.H. Bleijlevens, M.A. Lexis, S.M. Rasquin, J. Halfens, et al., *Using a Delphi Technique to Seek Consensus Regarding Definitions, Descriptions and Classification of Terms Related to Implicit and Explicit Forms of Motor Learning.* PLOS ONE, 2014. **9**(6): p. e100227.

218. Hall, D.A., H. Smith, E. Heffernan, K. Fackrell, and G. for the Core Outcome Measures in Tinnitus International Delphi Research Steering, *Recruiting and retaining participants in e-Delphi surveys for core outcome set development: Evaluating the COMiT'ID study.* PLOS ONE, 2018. **13**(7): p. e0201378.

219. Galloway, A.M., D.J. Keene, A. Anderson, C. Holton, A.C. Redmond, H.J. Siddle, et al., *Clinical consensus recommendations for the non-surgical treatment of children with Perthes' disease in the UK.* The Bone & Joint Journal, 2024. **106-B**(5): p. 501-507.

220. Funnell, S.C. and P.J. Rogers, *Purposeful program theory: Effective use of theories of change and logic models*. 2011: John Wiley & Sons.

221. Chen, K., *An Interactive Design Framework for Children's Apps for Enhancing Emotional Experience.* Interacting with Computers, 2022. **34**(3): p. 85-98.

222. Lee, J., Z. Su, and Y. Chen, *Mobile Apps for Children's Health and Wellbeing: Design Features and Future Opportunities.* AMIA Annu Symp Proc, 2023. **2023**: p. 1027-1036.

223. Excellence, N.I.f.H.a.C. *Evidence standards framework (ESF) for digital health technologies*. 2019 [cited 2024 15/05/2024]; Available from: https://www.nice.org.uk/Media/Default/About/what-we-do/our-programmes/evidence-standards-framework/digital-evidence-standards-framework.pdf.

224. Duncan, E., A. O'Cathain, N. Rousseau, L. Croot, K. Sworn, K.M. Turner, et al., *Guidance for reporting intervention development studies in health research (GUIDED): an evidence-based consensus study.* BMJ Open, 2020. **10**(4): p. e033516.

225. Rehfuess, E.A., A. Booth, L. Brereton, J. Burns, A. Gerhardus, K. Mozygemba, et al., *Towards a taxonomy of logic models in systematic reviews and health technology assessments: A priori, staged, and iterative approaches.* Res Synth Methods, 2018. **9**(1): p. 13-24.

226. Michie, S., M. Richardson, M. Johnston, C. Abraham, J. Francis, W. Hardeman, et al., *The Behavior Change Technique Taxonomy (v1) of 93 Hierarchically Clustered Techniques: Building an International Consensus for the Reporting of Behavior Change Interventions.* Annals of Behavioral Medicine, 2013. **46**(1): p. 81-95.

227. Maden, M., A. Cunliffe, N. McMahon, A. Booth, G.M. Carey, S. Paisley, et al., *Use of programme theory to understand the differential effects of interventions across socio-economic groups in systematic reviews—a systematic methodology review.* Systematic Reviews, 2017. **6**(1): p. 266.

228. O'Cathain, A., L. Croot, E. Duncan, N. Rousseau, K. Sworn, K.M. Turner, et al., *Guidance on how to develop complex interventions to improve health and healthcare.* BMJ Open, 2019. **9**(8): p. e029954.

229. Mills, T., R. Lawton, and L. Sheard, *Advancing complexity science in healthcare research: the logic of logic models.* BMC Medical Research Methodology, 2019. **19**(1): p. 55.

230. HMA. *DIGIBETE APP COMMISSIONED BY NHS ENGLAND AND NHS IMPROVEMENT*. 2024 [cited 2024 22/02/2024]; Available from: https://www.hma.co.uk/insights/digibete-app-commissioned-by-nhs-england-and-nhs-improvement/.

231. DigiBete. *DigiBete: Home page*. 2024 [cited 2024 22/02/2024]; Available from: https://www.digibete.org.

232. Chettri, S., V. Wang, E.A. Balkin, M.F. Rayo, and C.N. Lee, *Development of a Mobile App for Clinical Research: Challenges and Implications for Investigators.* JMIR Mhealth Uhealth, 2022. **10**(4): p. e32244.

233. Leeds, U.o. *Getting quotes and choosing a supplier*. 2024 [cited 2024 22/02/2024]; Available from: https://comms.leeds.ac.uk/creative-design-and-video/getting-quotes-and-choosing-your-supplier/.

234. Hernandez, H.A., Z. Ye, T.C.N. Graham, D. Fehlings, and L. Switzer, *Designing action-based exergames for children with cerebral palsy*, in *Proceedings of the SIGCHI Conference on Human Factors in Computing Systems*. 2013, Association for Computing Machinery: Paris, France. p. 1261–1270.

235. Balli, F., *Developing Digital Games to Address Airway Clearance Therapy in Children With Cystic Fibrosis: Participatory Design Process.* JMIR Serious Games, 2018. **6**(4): p. e18.

236. Garra, G., A.J. Singer, B.R. Taira, J. Chohan, H. Cardoz, E. Chisena, et al., *Validation of the Wong-Baker FACES Pain Rating Scale in pediatric emergency department patients.* Acad Emerg Med, 2010. **17**(1): p. 50-4.

237. Wong, D.L. and C.M. Baker, *Pain in children: comparison of assessment scales.* Pediatr Nurs, 1988. **14**(1): p. 9-17.

238. Garra, G., A.J. Singer, A. Domingo, and H.C. Thode, Jr., *The Wong-Baker pain FACES scale measures pain, not fear.* Pediatr Emerg Care, 2013. **29**(1): p. 17-20.

239. Varni, J.W., D. Thissen, B.D. Stucky, Y. Liu, H. Gorder, D.E. Irwin, et al., *PROMIS® Parent Proxy Report Scales: an item response theory analysis of the parent proxy report item banks.* Qual Life Res, 2012. **21**(7): p. 1223-40.

240. Varni, J.W., D. Thissen, B.D. Stucky, Y. Liu, B. Magnus, H. Quinn, et al., *PROMIS® Parent Proxy Report Scales for children ages 5-7 years: an item response theory analysis of differential item functioning across age groups.* Qual Life Res, 2014. **23**(1): p. 349-61.

241. Shields, B.J., D.M. Cohen, C. Harbeck-Weber, J.D. Powers, and G.A. Smith, *Pediatric pain measurement using a visual analogue scale: a comparison of two teaching methods.* Clin Pediatr (Phila), 2003. **42**(3): p. 227-34.

242. Perry, D.C., J. Achten, R. Knight, D. Appelbe, S.J. Dutton, M. Dritsaki, et al., *Immobilisation of torus fractures of the wrist in children (FORCE): a randomised controlled equivalence trial in the UK.* Lancet, 2022. **400**(10345): p. 39-47.

243. Jindal, P. and J.C. MacDermid, *Assessing reading levels of health information: uses and limitations of flesch formula.* Educ Health (Abingdon), 2017. **30**(1): p. 84-88.

244. Worldwide, S. *Perthes' Disease*. 2024 [cited 2024 15/05/2024]; Available from: https://www.stepsworldwide.org/conditions/perthes/.

245. Foundation, B.N. *Children*. 2024 [cited 2024 15/05/2024]; Available from: https://www.nutrition.org.uk/nutrition-for/children/.

246. Care, D.o.H.a.S. *UK Chief Medical Officers' Physical Activity Guidelines*. 2019 [cited 2024 15/05/2024]; Available from: https://assets.publishing.service.gov.uk/media/62063c6b8fa8f5109d2d6d53/physical-activity-for-children-and-young-people-5-to-18-years.pdf.

247. Trust, T.L.T.H.N. *Current studies: NON-STOP*. 2024 [cited 2024 15/05/2024]; Available from: https://www.leedsth.nhs.uk/services/childrens-clinical-research/current-studies/#non-stop.

248. matters, B.H.E.m. *Looking after a child or young person's mental health*. 2024 [cited 2024 15/05/2024]; Available from: https://www.nhs.uk/every-mind-matters/supporting-others/childrens-mental-health/.

249. Naeem, F. and S. Y, *Development, Testing and Reporting of Mobile Apps for Psycho-social Interventions: Lessons from the Pharmaceuticals.* Journal of Medical Diagnostic Methods, 2015. **04**.

250. Foster, M., *A Mobile Application for Patients With Heart Failure: Theory- and Evidence-Based Design and Testing.* CIN: Computers, Informatics, Nursing, 2018. **36**(11): p. 540-549.

251. Teo, C.H., C.J. Ng, S.K. Lo, C.D. Lim, and A. White, *A Mobile Web App to Improve Health Screening Uptake in Men (ScreenMen): Utility and Usability Evaluation Study.* JMIR Mhealth Uhealth, 2019. **7**(4): p. e10216.

252. UNICEF. *Closing the Digital Divide for Good: An end to the digital exclusion of children and young people in the UK*. 2022; Available from: https://www.unicef.org.uk/wp-content/uploads/2021/06/Closing-the-Digital-Divide-for-Good_FINAL.pdf.

253. Ofcom. *Children and parents: media use and attitudes report 2022*. 2022 [cited 2024 24/07/2024]; Available from: https://www.ofcom.org.uk/media-use-and-attitudes/media-habits-children/children-and-parents-media-use-and-attitudes-report-2022/.

254. Emerson, M.R., S. Buckland, M.A. Lawlor, D. Dinkel, D.J. Johnson, M.S. Mickles, et al., *Addressing and evaluating health literacy in mHealth: a scoping review.* Mhealth, 2022. **8**: p. 33.

255. Jang, Y. and B. Ko, *Online Safety for Children and Youth under the 4Cs Framework-A Focus on Digital Policies in Australia, Canada, and the UK.* Children (Basel), 2023. **10**(8).

256. Evans, S.R., *Clinical trial structures.* J Exp Stroke Transl Med, 2010. **3**(1): p. 8-18.

257. Heary, C.M. and E. Hennessy, *The use of focus group interviews in pediatric health care research.* J Pediatr Psychol, 2002. **27**(1): p. 47-57.

258. Hildebrand, D.L., *Pragmatic democracy: Inquiry, objectivity, and experience.* Metaphilosophy, 2011. **42**(5): p. 589-604.

259. Ji, Z., J. Lin, and J. Lin, *Optimal sample size determination for single-arm trials in pediatric and rare populations with Bayesian borrowing.* Journal of Biopharmaceutical Statistics, 2022. **32**(4): p. 529-546.

260. Julious, S.A., *Sample sizes for clinical trials*. 2023: chapman and hall/CRC.

261. Hennink, M.M., B.N. Kaiser, and M.B. Weber, *What Influences Saturation? Estimating Sample Sizes in Focus Group Research.* Qual Health Res, 2019. **29**(10): p. 1483-1496.

262. Adler, K., S. Salanterä, and M. Zumstein-Shaha, *Focus Group Interviews in Child, Youth, and Parent Research: An Integrative Literature Review.* International Journal of Qualitative Methods, 2019. **18**: p. 1609406919887274.

263. Perry, D.C. and A.J. Hall, *The Epidemiology and Etiology of Perthes Disease.* Orthopedic Clinics of North America, 2011. **42**(3): p. 279-283.

264. Statistics, O.f.N. *Measuring equality: A guide for the collection and classification of ethnic group, national identity and religion data in the UK*. 2021 [cited 2024 21/08/2024]; Available from: https://www.ons.gov.uk/methodology/classificationsandstandards/measuringequality/ethnicgroupnationalidentityandreligion.

265. Eldridge, S.M., C.L. Chan, M.J. Campbell, C.M. Bond, S. Hopewell, L. Thabane, et al., *CONSORT 2010 statement: extension to randomised pilot and feasibility trials.* Bmj, 2016. **355**: p. i5239.

266. Centre, N.C.B.R. *Children's Physical Activity Questionnaire*. 2017 [cited 2023 21/07/2023]; Available from: https://www.measurement-toolkit.org/instrument/37#:~:text=The%20Child%20Physical%20Activity%20Questionnaire,over%20the%20past%207%20days.&text=Frequency%20and%20duration%20of%20a,for%20weekdays%20and%20weekend%20days.

267. Corder, K., E.M. van Sluijs, A. Wright, P. Whincup, N.J. Wareham, and U. Ekelund, *Is it possible to assess free-living physical activity and energy expenditure in young people by self-report?* Am J Clin Nutr, 2009. **89**(3): p. 862-70.

268. Schnall, R., H. Cho, and J. Liu, *Health Information Technology Usability Evaluation Scale (Health-ITUES) for Usability Assessment of Mobile Health Technology: Validation Study.* JMIR Mhealth Uhealth, 2018. **6**(1): p. e4.

269. Leo, D.G., D.C. Perry, B. Abdullah, and H. Jones, *PROMIS Paediatric Mobility tool is correlated with accelerometer-measured physical activity in children with hip diseases.* The Bone & Joint Journal, 2021. **103-B**(2): p. 405-410.

270. Measures, P.H. *Obtain & Administer Measures*. 2024 [cited 2024 01/11/2024]; Available from: https://www.healthmeasures.net/explore-measurement-systems/promis/obtain-administer-measures#:~:text=All%20English%20and%20some%20Spanish,without%20licensing%20or%20royalty%20fees.

271. Anderson, Y.C., L.E. Wynter, C.C. Grant, J.M. Stewart, T.L. Cave, C.E. Wild, et al., *Physical activity is low in obese New Zealand children and adolescents.* Sci Rep, 2017. **7**: p. 41822.

272. Harrell, J.S., R.G. McMurray, C.D. Baggett, M.L. Pennell, P.F. Pearce, and S.I. Bangdiwala, *Energy costs of physical activities in children and adolescents.* Med Sci Sports Exerc, 2005. **37**(2): p. 329-36.

273. Yen, P.Y., K.H. Sousa, and S. Bakken, *Examining construct and predictive validity of the Health-IT Usability Evaluation Scale: confirmatory factor analysis and structural equation modeling results.* J Am Med Inform Assoc, 2014. **21**(e2): p. e241-8.

274. Yen, P.Y., D. Wantland, and S. Bakken, *Development of a Customizable Health IT Usability Evaluation Scale.* AMIA Annu Symp Proc, 2010. **2010**: p. 917-21.

275. Loh, K.P., J. Liu, S. Ganzhorn, G. Sanabria, and R. Schnall, *Establishing a usability cut-point for the health information technology usability evaluation scale (Health-ITUES).* Int J Med Inform, 2022. **160**: p. 104713.

276. Loh, K.P., J. Liu, S. Ganzhorn, G. Sanabria, and R. Schnall, *Establishing a usability cut-point for the health information technology usability evaluation scale (Health-ITUES).* International Journal of Medical Informatics, 2022. **160**: p. 104713.

277. von Rosing, M., U. Foldager, M. Hove, J. von Scheel, and A.F. Bøgebjerg, *Working with the Business Process Management (BPM) Life Cycle*, in *The Complete Business Process Handbook*, M. von Rosing, A.-W. Scheer, and H. von Scheel, Editors. 2015, Morgan Kaufmann: Boston. p. 269-345.

278. Cotterill, S., S. Knowles, A.M. Martindale, R. Elvey, S. Howard, N. Coupe, et al., *Getting messier with TIDieR: embracing context and complexity in intervention reporting.* BMC Med Res Methodol, 2018. **18**(1): p. 12.

279. Hoffmann, T.C., P.P. Glasziou, I. Boutron, R. Milne, R. Perera, D. Moher, et al., *Better reporting of interventions: template for intervention description and replication (TIDieR) checklist and guide.* BMJ : British Medical Journal, 2014. **348**: p. g1687.

280. Wong, L.P., *Focus group discussion: a tool for health and medical research.* Singapore Med J, 2008. **49**(3): p. 256-60; quiz 261.

281. Santhosh, L., J.C. Rojas, and P.G. Lyons, *Zooming into Focus Groups: Strategies for Qualitative Research in the Era of Social Distancing.* ATS Sch, 2021. **2**(2): p. 176-184.

282. Phillippi, J. and J. Lauderdale, *A Guide to Field Notes for Qualitative Research: Context and Conversation.* Qualitative Health Research, 2018. **28**(3): p. 381-388.

283. Tong, A., P. Sainsbury, and J. Craig, *Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups.* International Journal for Quality in Health Care, 2007. **19**(6): p. 349-357.

284. Pupovac, V. and M. Petrovecki, *Summarizing and presenting numerical data.* Biochem Med (Zagreb), 2011. **21**(2): p. 106-10.

285. Borrelli, B., *The Assessment, Monitoring, and Enhancement of Treatment Fidelity In Public Health Clinical Trials.* J Public Health Dent, 2011. **71**(s1): p. S52-s63.

286. Lancaster, K., A. Bhopti, M.L. Kern, R. Taylor, A. Janson, and K. Harding, *Effectiveness of peer support programmes for improving well-being and quality of life in parents/carers of children with disability or chronic illness: A systematic review.* Child: Care, Health and Development, 2023. **49**(3): p. 485-496.

287. Abdelazeem, B., K.S. Abbas, M.A. Amin, N.A. El-Shahat, B. Malik, A. Kalantary, et al., *The effectiveness of incentives for research participation: A systematic*

review and meta-analysis of randomized controlled trials. PLoS One, 2022. **17**(4): p. e0267534.

288. Panagioti, M., G. Richardson, N. Small, E. Murray, A. Rogers, A. Kennedy, et al., *Self-management support interventions to reduce health care utilisation without compromising outcomes: a systematic review and meta-analysis.* BMC Health Serv Res, 2014. **14**: p. 356.
289. Lindsay, S., S. Kingsnorth, C. McDougall, and H. Keating, *A systematic review of self-management interventions for children and youth with physical disabilities.* Disabil Rehabil, 2014. **36**(4): p. 276-88.
290. Brigden, A., E. Anderson, C. Linney, R. Morris, R. Parslow, T. Serafimova, et al., *Digital Behavior Change Interventions for Younger Children With Chronic Health Conditions: Systematic Review.* J Med Internet Res, 2020. **22**(7): p. e16924.
291. Charlier, N., N. Zupancic, S. Fieuws, K. Denhaerynck, B. Zaman, and P. Moons, *Serious games for improving knowledge and self-management in young people with chronic conditions: a systematic review and meta-analysis.* J Am Med Inform Assoc, 2016. **23**(1): p. 230-9.
292. Kumar, V.S., K.J. Wentzell, T. Mikkelsen, A. Pentland, and L.M. Laffel, *The DAILY (Daily Automated Intensive Log for Youth) trial: a wireless, portable system to improve adherence and glycemic control in youth with diabetes.* Diabetes Technol Ther, 2004. **6**(4): p. 445-53.
293. Gkintoni, E., F. Vantaraki, C. Skoulidi, P. Anastassopoulos, and A. Vantarakis, *Promoting Physical and Mental Health among Children and Adolescents via Gamification-A Conceptual Systematic Review.* Behav Sci (Basel), 2024. **14**(2).
294. Modi, A.C., A.L. Pai, K.A. Hommel, K.K. Hood, S. Cortina, M.E. Hilliard, et al., *Pediatric self-management: a framework for research, practice, and policy.* Pediatrics, 2012. **129**(2): p. e473-85.
295. Zullig, L.L., C. Drake, D.K. Check, T. Brunkert, M. Deschodt, M. Olson, et al., *Embedding implementation science in the research pipeline.* Translational Behavioral Medicine, 2023. **14**(2): p. 73-79.

296. Library, N.A. *The NHS Apps Library has closed*. 2021 [cited 2024 14/10/2024]; Available from: https://www.nhs.uk/apps-library/.

297. Klaic, M., S. Kapp, P. Hudson, W. Chapman, L. Denehy, D. Story, et al., *Implementability of healthcare interventions: an overview of reviews and development of a conceptual framework.* Implementation Science, 2022. **17**(1): p. 10.

298. England, N. *Digital Technology Assessment Criteria (DTAC)*. 2021 [cited 2024 14/10/2024]; Available from: https://transform.england.nhs.uk/key-tools-and-info/digital-technology-assessment-criteria-dtac/.

299. England, N. *The Digital Technology Assessment Criteria for Health and Social Care (DTAC)*. 2021 [cited 2024 14/10/2024]; Available from: https://view.officeapps.live.com/op/view.aspx?src=https%3A%2F%2Ftransform.england.nhs.uk%2Fmedia%2Fdocuments%2FDTAC_version_1.0_FINAL_updated_16.04.odt&wdOrigin=BROWSELINK.

300. Sweet, S.N., K.A.M. Ginis, P.A. Estabrooks, and A.E. Latimer-Cheung, *Operationalizing the RE-AIM framework to evaluate the impact of multi-sector partnerships.* Implementation Science, 2014. **9**(1): p. 74.

301. Briatico, D., K.C. Reilly, P. Tucker, J.D. Irwin, A.M. Johnson, E.S. Pearson, et al., *Using the RE-AIM framework to evaluate the feasibility of a parent-focused intervention targeting childhood obesity.* Pilot and Feasibility Studies, 2023. **9**(1): p. 38.

302. Burke, S.M., S. Shapiro, R.J. Petrella, J.D. Irwin, M. Jackman, E.S. Pearson, et al., *Using the RE-AIM framework to evaluate a community-based summer camp for children with obesity: a prospective feasibility study.* BMC obesity, 2015. **2**: p. 1-10.

303. Burke, S.M., L.M. Vanderloo, A. Gaston, E.S. Pearson, and P. Tucker, *An Examination of Self-Reported Physical Activity and Physical Activity Self-Efficacy Among Children with Obesity: Findings from the Children's Health and Activity Modification Program (C.H.A.M.P.) Pilot Study (Análisis de la actividad física autoreportad.* Retos, 2015. **28**(0): p. 212-218.

304. Holtrop, J.S., P.A. Estabrooks, B. Gaglio, S.M. Harden, R.S. Kessler, D.K. King, et al., *Understanding and applying the RE-AIM framework: Clarifications and resources.* J Clin Transl Sci, 2021. **5**(1): p. e126.

305. Willmott, T.J., B. Pang, and S. Rundle-Thiele, *Capability, opportunity, and motivation: an across contexts empirical examination of the COM-B model.* BMC Public Health, 2021. **21**(1): p. 1014.

306. McQuinn, S., S. Belton, A. Staines, and M.R. Sweeney, *Co-design of a school-based physical activity intervention for adolescent females in a disadvantaged community: insights from the Girls Active Project (GAP).* BMC Public Health, 2022. **22**(1): p. 615.

307. Birken, S.A., C.L. Rohweder, B.J. Powell, C.M. Shea, J. Scott, J. Leeman, et al., *T-CaST: an implementation theory comparison and selection tool.* Implement Sci, 2018. **13**(1): p. 143.

308. Birken SA, R.C., Powell BJ, Shea CM, Scott J, Leeman J, Grewe ME, Alexis Kirk M, Damschroder L, Aldridge WA 2nd, Haines ER, Straus S, Presseau J. *Theory, Model, and Framework Comparison and Selection Tool (T-CaST) for Implementation Researchers*. 2018 [cited 2024 15/10/2024]; Available from: https://impsci.tracs.unc.edu/wp-content/uploads/T-CaST_researchers.pdf.

309. Statista. *Number of apps available in leading app stores as of August 2024*. 2024 [cited 2024 10/10/2024]; Available from: https://www.statista.com/statistics/276623/number-of-apps-available-in-leading-app-stores/#:~:text=Number%20of%20apps%20available%20in%20leading%20app%20stores%202024&text=As%20of%20August%202024%2C%20Android,million%20available%20apps%20for%20iOS.

310. Anderson, A.M., G.A. McHugh, C. Comer, J. Joseph, T.O. Smith, L. Yardley, et al., *Supporting patients to prepare for total knee replacement: Evidence-, theory- and person-based development of a 'Virtual Knee School' digital intervention.* Health Expect, 2023. **26**(6): p. 2549-2570.

311. Griffin, D.R., E.J. Dickenson, P.D.H. Wall, F. Achana, J.L. Donovan, J. Griffin, et al., *Hip arthroscopy versus best conservative care for the treatment of*

femoroacetabular impingement syndrome (UK FASHIoN): a multicentre randomised controlled trial. The Lancet, 2018. **391**(10136): p. 2225-2235.

312. Keene, D.J., M.L. Costa, N. Peckham, E. Tutton, V.S. Barber, S.J. Dutton, et al., *Progressive exercise versus best practice advice for adults aged 50 years or over after ankle fracture: the AFTER pilot randomised controlled trial.* BMJ Open, 2022. **12**(11): p. e059235.
313. Fàbregues, S., C. Mumbardó-Adam, E.L. Escalante-Barrios, Q.N. Hong, D. Edelstein, K. Vanderboll, et al., *Mixed methods intervention studies in children and adolescents with emotional and behavioral disorders: A methodological review.* Research in Developmental Disabilities, 2022. **126**: p. 104239.
314. Research, N.I.f.H.a.C. *Participant in Research Experience Survey (PRES)*. 2024 [cited 2024 21/10/2024]; Available from: https://www.nihr.ac.uk/about-us/who-we-are/reports-and-performance/participant-in-research-experience-survey.
315. Group, P.S. *Public Involvement Impact Assessment Framework (PiiAF).* 2014 [cited 2024 21/10/2024]; Available from: https://piiaf.org.uk/index.php.
316. Collins, M., R. Long, A. Page, J. Popay, and F. Lobban, *Using the Public Involvement Impact Assessment Framework to assess the impact of public involvement in a mental health research context: A reflective case study.* Health Expect, 2018. **21**(6): p. 950-963.
317. Staniszewska, S., J. Brett, I. Simera, K. Seers, C. Mockford, S. Goodlad, et al., *GRIPP2 reporting checklists: tools to improve reporting of patient and public involvement in research.* BMJ, 2017. **358**: p. j3453.
318. Bauer, M.S., L. Damschroder, H. Hagedorn, J. Smith, and A.M. Kilbourne, *An introduction to implementation science for the non-specialist.* BMC Psychol, 2015. **3**(1): p. 32.
319. Ramly, E. and H.W. Brown, *Beyond Effectiveness: Implementation Science 101 for Clinicians and Clinical Researchers.* Urogynecology (Phila), 2023. **29**(3): p. 307-312.

320. Sarkies, M.N., E. Francis-Auton, J.C. Long, C. Pomare, R. Hardwick, and J. Braithwaite, *Making implementation science more real.* BMC Medical Research Methodology, 2022. **22**(1): p. 178.

321. X, B.a.J.J.v. *Most talked about, May 2024*. 2024 [cited 2024 03/12/2024]; Available from: https://x.com/BoneJointJ/status/1803306865234673681.

322. TOTS. *Trauma & Orthopaedic Trials for Children*. 2024 [cited 2024 16/10/2024]; Available from: https://tots.digitrial.com.

323. ROBUST. *The ROBUST Study: Strengthening programme for ambulant adolescents with cerebral palsy*. 2024 [cited 2024 21/10/2024]; Available from: https://robust-study.org.

324. SPELL. *The SPELL study: Stretching programme for ambulant adolescents with cerebral palsy*. 2024 [cited 2024 21/10/2024]; Available from: https://www.spell-study.org.

325. Research, N.I.f.H.a.C. *Best Research for Best Health: The Next Chapter*. 2021 [cited 2024 03/12/2024]; Available from: https://www.nihr.ac.uk/best-research-best-health-next-chapter#tab-106656.

326. Beni, R., S.A. Hussain, F. Monsell, and Y. Gelfer, *Management of Legg-Calve-Perthes disease: a scoping review with advice on initial management.* Archives of Disease in Childhood, 2024: p. archdischild-2024-327891.

327. Bijker, L., L. de Wit, P. Cuijpers, E. Poolman, G. Scholten-Peeters, and M.W. Coppieters, *Back2Action: effectiveness of physiotherapy blended with eHealth consisting of pain education and behavioural activation versus physiotherapy alone—protocol for a pragmatic randomised clinical trial for people with subacute or persistent spinal pain.* BMJ Open, 2022. **12**(1): p. e050808.

328. Murray, E., E.B. Hekler, G. Andersson, L.M. Collins, A. Doherty, C. Hollis, et al., *Evaluating Digital Health Interventions: Key Questions and Approaches.* Am J Prev Med, 2016. **51**(5): p. 843-851.

329. Ginsburg, L.R., M. Hoben, A. Easterbrook, R.A. Anderson, C.A. Estabrooks, and P.G. Norton, *Fidelity is not easy! Challenges and guidelines for assessing fidelity in complex interventions.* Trials, 2021. **22**(1): p. 372.

330. registry, B.I. *Operative or non-surgical treatment of Perthes' Disease*. 2024 [cited 2024 03/12/2024]; Available from: https://www.isrctn.com/ISRCTN83315571.

331. Research, N.I.f.H.a.C. *Op Non-STOP Study (Operative or Non-Surgical Treatment of Perthes' Disease) - A multi-centre prospective randomised superiority trial of containment surgery compared to optimised non-surgical care for Perthes' disease of the hip in children*. 2024 [cited 2024 24/11/2024]; Available from: https://fundingawards.nihr.ac.uk/award/NIHR152309.

332. Galloway, A.M., N. Nicolaou, and D.C. Perry, *Surgery versus the non-surgical treatment of Perthes' disease (Op Non-STOP).* The Bone & Joint Journal, 2025. **107-B**(3): p. 280-282.

Appendices

Appendix A – PROMIS Mobility

PROMIS® Pediatric Item Bank GenPop v3.0 – Mobility

Pediatric Mobility

Please respond to each question or statement by marking one box per row.

In the past 7 days...

		With no trouble	With a little trouble	With some trouble	With a lot of trouble	Not able to do
4124R1r2	I could get up from the floor.....	☐ 4	☐ 3	☐ 2	☐ 1	☐ 1
236R1r2	I could keep up when I played with other kids	☐ 4	☐ 3	☐ 2	☐ 1	☐ 1
3892R1r2	I could move my legs	☐ 4	☐ 3	☐ 2	☐ 1	☐ 1
2646R1r2	I could stand up by myself.....	☐ 4	☐ 3	☐ 2	☐ 1	☐ 1
4185R1r2	I could stand up on my tiptoes.....	☐ 4	☐ 3	☐ 2	☐ 1	☐ 1
2707R2r2	I could walk up stairs without holding on to anything	☐ 4	☐ 3	☐ 2	☐ 1	☐ 1
5023R1r2	I have been physically able to do the activities I enjoy most.....	☐ 4	☐ 3	☐ 2	☐ 1	☐ 1
2117R1r	I could ride a bike	☐ 5	☐ 4	☐ 3	☐ 2	☐ 1
2118R1r2	I could get in and out of a car	☐ 4	☐ 3	☐ 2	☐ 1	☐ 1
219R1r2	I could walk more than one block	☐ 4	☐ 3	☐ 2	☐ 1	☐ 1

ITEM BANKS ARE NOT INTENDED TO BE ADMINISTERED IN THEIR ENTIRETY.

12 May 2023
© 2010-2023 PROMIS Health Organization (PHO)

In the past 7 days...

		With no trouble	With a little trouble	With some trouble	With a lot of trouble	Not able to do
2202R2r	I could walk across the room.....	☐ 4	☐ 3	☐ 2	☐ 1	☐ 1
2642aR1r2	I could get out of bed by myself.....	☐ 4	☐ 3	☐ 2	☐ 1	☐ 1
2642bR1r2	I could get into bed by myself.....	☐ 4	☐ 3	☐ 2	☐ 1	☐ 1
2647R2r2	I could get down on my knees without holding on to something.....	☐ 4	☐ 3	☐ 2	☐ 1	☐ 1
3799R1r2	I could carry my books in my backpack...	☐ 4	☐ 3	☐ 2	☐ 1	☐ 1
4079R1r2	I could get up from a regular toilet.....	☐ 4	☐ 3	☐ 2	☐ 1	☐ 1
4137R1r2	I could go up one step.....	☐ 4	☐ 3	☐ 2	☐ 1	☐ 1
4190R1r2	I could turn my head all the way to the side.....	☐ 4	☐ 3	☐ 2	☐ 1	☐ 1
5200bR1r	I could run a mile.....	☐ 5	☐ 4	☐ 3	☐ 2	☐ 1
676R1r2	I could bend over to pick something up ...	☐ 4	☐ 3	☐ 2	☐ 1	☐ 1
7026r	I could jump up and down.....	☐ 4	☐ 3	☐ 2	☐ 1	☐ 1

ITEM BANKS ARE NOT INTENDED TO BE ADMINISTERED IN THEIR ENTIRETY.

© 2010-2023 PROMIS Health Organization (PHO)

Appendix B – Child participant information sheet

UNIVERSITY OF LEEDS

NON-STOP: NON-Surgical Treatment Of Perthes' Disease

Chief Investigator(s):

Professor Anthony Redmond and Mr Adam Galloway

INFORMATION SHEET FOR CHILDREN

BACKGROUND

The NON-STOP study is looking at how we look after children with Perthes' Disease.

You and your guardian/carer said that we could talk to you about your Perthes' Disease.

We want to know what you have liked about your treatment and what you have not like as much. We also want to tell you about a new app we're making for people with Perthes' Disease.

There are a few more things we need to tell you.

DO I HAVE TO SAY YES?

No. If you say no, that's okay and it won't change how people in the hospital look after you.

WHAT HAPPENS IF I SAY YES?

We will ask you to say that it is okay for us to talk to you on a phone call, this might be on a video call like a FaceTime or we can do it in person.

I will ask your guardian/carer to say it is okay to talk on the phone with me, or write their name on a form.

You can ask any questions about the study.

We will ask you some questions about what types of things people in the hospital normally ask you to do for your Perthes' Disease. We'll record the interviews so we can remember them later.

The interview will take about 20 minutes for us to talk, but you can stop whenever you want.

Your guardian/carer will be with you the whole time.

WHO WILL KNOW WHO I AM IN THE STUDY?

Only people doing the research will know who you are in the study.

WHAT HAPPENS IF I WANT TO STOP?

You can stop talking whenever you want to, and we will stop.

Thank you

Appendix C – Family participant information sheet

UNIVERSITY OF LEEDS

NON-STOP: NON-Surgical Treatment Of Perthes' Disease

Chief Investigator(s):

Professor Anthony Redmond and Mr Adam Galloway

INFORMATION SHEET FOR PARENT/GUARDIAN

BACKGROUND

The NON-STOP study is aiming to improve how much we understand about the care that our patients and their families receive for Perthes' Disease. Previously you agreed we could contact you to discuss taking part in an interview with one of our researchers. The aim of the interview is to discuss what your experiences of care have been for Perthes' Disease as well as discuss what future care might look like.

This document will explain in more detail what the study will include. **Please take time to read this and feel free to discuss with others if you wish.**

WHAT HAPPENS IF I SAY YES?

If you decide to take part in the interview, we will ask you to reply to the email that contained this sheet, with an 'agreement statement' as described, this will act as consent to take part in the study. It will say that you have read this information sheet, had time to decide whether to be involved and agree to take part. We will then arrange a date and time for your interview. You can choose to be interviewed using either a video call or a normal phone call or, if you would prefer it, we can complete the interview in person, either in the hospital you normally go to, or in your home.

The interview will be with you and your child, and will take been 30-90 minutes, we will likely only need to speak to your child for around 20 minutes and you will be with them the whole time. If done using a phone call or video call the interview will be recorded using the same device. If done in person it will be recorded using a small recording device so that the researcher can listen to your answers again at a later time.

During the interview we will ask you both a range of questions about the care you have received for Perthes' Disease and talk to you both about what treatment for the condition might look like in the future.

DO I HAVE TO TAKE PART?

No. It is completely up to you whether you would like to take part, and you do not need to decide straight away. You are also free to change your mind and withdraw from the study at any point. It is also absolutely fine if your child doesn't want to take part, we can still interview you. Whether you/your child decide to take part or not will not affect the clinical care that your child receives.

UNIVERSITY OF LEEDS

ARE THERE ANY BENEFITS TO TAKING PART?

There are no specific benefits to taking part in the interview. However sometimes people feel there is a benefit to sharing their experiences and feel that being involved in research studies like this can help contribute to a better understanding of the condition. This might be particularly important in a condition like Perthes' Disease where we don't know an awful lot about the experiences of those most involved.

ARE THERE ANY RISKS IN TAKING PART?

Similar to the benefits, there are no specific risks in taking part. It is possible that having to recall previous experiences could bring back memories of something potentially upsetting. The research team will be able provide support however, and offer the chance to take a break or, if needed, stop the interview at any point.

In the rare occasion there are any safeguarding issues, confidential information may need to be shared with your existing medical team.

We appreciate that it can be time-consuming to take part in research, and because of this, we have funding to reimburse you and your child for your time and have a childcare allowance if this makes it more realistic for you to take part. In line with NIHR guidance this will be a maximum of £50.

WILL MY INFORMATION BE KEPT CONFIDENTIAL?

Yes. Your contact details [name, email address and phone number] will be kept in password-protected files. Only the research study team will have access to this. After the study you will be sent a summary of the research study and then your details will be destroyed. A company affiliated with the university will type up the recording of the interview (removing names/details that might identify your family) and we will keep these for ten years after the study has finished. This is in line with the laws on doing research, after this, they will be destroyed as well. We might use direct quotes from what you say in the interview but we won't name you or make it so anyone would know it was you who said it.

If you have any concerns about data privacy during the study you can email dpo@leeds.ac.uk

Alternatively, you can visit https://dataprotection.leeds.ac.uk/wp-content/uploads/sites/48/2020/08/My_data_and_research.pdf or https://dataprotection.leeds.ac.uk/wp-content/uploads/sites/48/2019/02/Research-Privacy-Notice.pdf or HRA website www.hra.nhs.uk/information-about-patients/

for more information on data privacy.

UNIVERSITY OF LEEDS

WHAT WILL HAPPEN TO THE RESULTS OF THE STUDY?

As mentioned previously, you will receive a summary of the study once we have analysed the interviews. There will be reports published in medical journals and at conferences, that will be available to you if you wish, but these will not name any participants. We will also provide a summary of the research findings on social media pages and relevant Perthes' Disease charity pages.

This piece of research is being done as part of a PhD; the results will contribute to the thesis written as part of this.

WHO HAS REVIEWED THIS STUDY?

Every piece of research that takes place in the NHS is reviewed by a group of specialists called a Research Ethics Committee. This is to make sure that any research conducted is done with the least risk of burden possible for the participants. This study has been reviewed by NHS West of Scotland Research and Ethics Committee 1 and deemed safe to proceed.

WHAT IF THERE IS A PROBLEM?

This study is sponsored by the University of Leeds. If you wish to discuss any aspect of the research study then you can contact the Chief Investigator, Professor Anthony Redmond at a.redmond@leeds.ac.uk.

If there are any issues/concerns that you wish to discuss that are about your clinical care, please discuss this with the interviewer who will inform your doctor at the relevant hospital. They will be able to put you in contact with the local Patient Advice and Liaison Service.

If you wish to withdraw at any point during the study, we can destroy the information you have provided.

FURTHER INFORMATION AND CONTACT DETAILS

If there are any other questions or concerns about the study or the interviews, please contact Adam Galloway at a.galloway@leeds.ac.uk

Thank you for taking the time to read this information and consider taking part in our study.

Appendix D – Clinician participant information sheet

UNIVERSITY OF LEEDS

NON-STOP: NON-Surgical Treatment Of Perthes' Disease

Chief Investigator(s):

Professor Anthony Redmond and Mr Adam Galloway

INFORMATION SHEET FOR CLINICIANS

BACKGROUND

The NON-STOP study is aiming to improve how much we understand the care that our patients and their families receive for Perthes' Disease. Previously you agreed we could contact you to discuss taking part in an interview with one of our researchers. The aim of the interview is to understand the information and experiences that inform the decision making of clinicians who regularly manage children with Perthes' Disease and to identify the barriers and enablers to providing non-surgical care. We will also explore the perception of all participants towards the development of a digital intervention to support best non-surgical treatment of Perthes' Disease.

This document will explain in more detail what the study will include. **Please take time to read this and feel free to discuss with others if you wish.**

WHAT HAPPENS IF I SAY YES?

If you decide to take part in the interview, we will ask you to reply to the email that contained this information sheet with an 'agreement statement' as described. This will act as consent to take part in the study. It will say that you have read this information sheet, had time to decide whether to be involved and agree to take part. We will then arrange a date and time for your interview. This will take place using either a video call or a normal phone call and will be your choice.

The interview will take around 60 minutes. It will be recorded using the same system as the call, for example, if on video, the video call audio will be recorded.

During the interview we will aim to ask you a range of questions about your experiences of providing care for this patient group as well as the digital intervention.

If you have any concerns about data privacy during the study you can email dpo@leeds.ac.uk

Alternatively, you can visit https://dataprotection.leeds.ac.uk/wp-content/uploads/sites/48/2020/08/My_data_and_research.pdf or https://dataprotection.leeds.ac.uk/wp-content/uploads/sites/48/2019/02/Research-Privacy-Notice.pdf or HRA website www.hra.nhs.uk/information-about-patients/ for more information on data privacy.

UNIVERSITY OF LEEDS

DO I HAVE TO TAKE PART?

No. It is completely up to you if you would like to take part, and you do not need to decide straight away. You are also free to change your mind and withdraw from the study at any point as well, without giving a reason. Your clinical service will not be informed of your participation.

ARE THERE ANY BENEFITS TO TAKING PART?

There are no specific benefits to taking part in the interview. However sometimes people feel there is a benefit to sharing their experiences and feel that being involved in research studies like this can help contribute to a better understanding of the condition. Particularly in a condition like Perthes' Disease where we don't know an awful lot about the experiences of those most involved.

ARE THERE ANY RISKS TO TAKING PART?

Similar to the benefits, there are no identified risks to taking part.

WILL MY INFORMATION BE KEPT CONFIDENTIAL?

Yes. Your contact details [name, email address and phone number] will be kept in password-protected files until the study has finished. At this point you will be sent a summary of the research study and then your details will be destroyed. A company affiliated with the university will type up the recording of the interview and we will keep these for ten years after the study has finished. This is in line with the laws on doing research, after this, they will be destroyed as well. We might use direct quotes from your interview but you will not be identifiable.

WHAT WILL HAPPEN TO THE RESULTS OF THE STUDY?

As mentioned previously, you will receive a summary of the study once we have analysed the interviews. There will be reports published in medical journals and at conferences, but these will not name any participants. We will also provide a summary of the research findings on social media pages and relevant Perthes' Disease charity pages.

This piece of research is being done as part of a PhD; the results will contribute to the thesis written as part of this.

WHO HAS REVIEWED THIS STUDY?

Every piece of research that takes place in the NHS is reviewed by a group of specialists called a Research Ethics Committee. This is to make sure that any research conducted is done with the least risk of burden possible to the participants. This study has been reviewed by NHS West of Scotland Research and Ethics Committee 1 and deemed safe to proceed.

WHAT IF THERE IS A PROBLEM?

This study is sponsored by the University of Leeds. If you wish to discuss any aspect of the research study then you can contact the Chief Investigator, Professor Anthony Redmond at a.redmond@leeds.ac.uk.

UNIVERSITY OF LEEDS

If there are any issues/concerns that you wish to discuss that are about your clinical care, please discuss this with the interviewer who will inform your doctor at the relevant hospital. They will be able to put you in contact with the local Patient Advice and Liaison Service.

FURTHER INFORMATION AND CONTACT DETAILS

If there are any other questions or concerns about the study or the interviews, please contact Adam Galloway at a.galloway@leeds.ac.uk

Thank you for taking the time to read this information and consider taking part in our study.

Appendix E – Topic guide for qualitative study

NON-STOP: NON-Surgical Treatment Of Perthes' Disease

Chief Investigator(s):

Professor Anthony Redmond and Mr Adam Galloway

TOPIC GUIDE FOR CHILD/FAMILY

AIM OF THIS GUIDE

This guide is for the interviewer to use in order to guide the interview process with the intended participant in this instance, the child/family dyad (pair).

It includes some questions and prompts that can be used during the interview as well as reminders for the interviewer as to the format of the interview.

IMPORTANT STEPS PRIOR TO STARTING INTERVIEW

Ensure participants have had time to read and understand the participant information pack and any further check for any further questions they may have.

Ensure participants understand that they do not have to take part in the interview and that they can stop the interview at any time.

At the beginning of the interview, make it clear that the interview will be recorded but that everything said will remain confidential and any information used will be anonymised. Their clinical care team will not be informed about any of the answers that they give in the interview. Be sure to inform the participant that recording has started and stopped.

QUESTIONS/PROMPTS TO BE USED DURING INTERVIEW

Child:

- What do you know about your hip problem (Perthes' Disease)?
- What treatment do you do for your hip? If physio, what physio?
- What do you like/not like about the treatment of your hip problem? Why?
    - Do you use any apps on your phone/tablet at the moment? If not, how would you use an app?
    - How do you feel about using an app to help you with Perthes' Disease?

Parent/legal guardian:

- What are your experiences of treatment of Perthes' Disease?
- What are your thoughts on non-surgical treatment of Perthes' Disease?
- What works well/not so well in terms of treatment of Perthes' Disease?
- Can you tell me about a time you were given a choice about your child's treatment for Perthes' Disease?
- What are your thoughts on using an app to help with your child's management of Perthes' Disease?
    - What sort of things would an app like this include?
- Ask child/family if they have any questions or if there is something else that they would like to add.

FINAL ACTIONS

Reiterate the plan following interview i.e. interviews with other participants, data analysis and the dissemination plan.

"How have you found the interview" – as a transition 'out' of the interview.

NON-STOP: NON-Surgical Treatment Of Perthes' Disease

Chief Investigator(s):

Professor Anthony Redmond and Mr Adam Galloway

TOPIC GUIDE FOR CLINICIANS

AIM OF THIS GUIDE

This guide is for the interviewer to use in order to guide the interview process with the intended participant in this instance, the clinician.

It includes some questions and prompts that can be used during the interview as well as reminders for the interviewer as to the format of the interview.

IMPORTANT STEPS PRIOR TO STARTING INTERVIEW

Ensure that participants understand that they do not have to take part in the interview.

Ensure that participants understand that they can stop the interview at any time.

At the beginning of the interview, make it clear that the interview will be recorded but that everything said will remain confidential and any information used will be anonymised. Be sure to inform the participant that recording has started and stopped.

Their clinical team will not be informed of any answers given during this interview.

QUESTIONS/PROMPTS TO BE USED DURING INTERVIEW

- What is your experience of treatment of Perthes' Disease?
  - If particularly successful/unsuccessful, why do you think this is?
- What is your experience of non-surgical treatment of Perthes' Disease?
  - If particularly successful/unsuccessful, why do you think this is?
- What are the factors when considering non-surgical treatment of Perthes' Disease?
  - Any barriers or enablers to making this decision?
  - What are the key influences?
- What is important to you in terms of treatment of Perthes' Disease?
  - What works well/not so well?
- What are your thoughts on using an app to help with management of Perthes' Disease?
  - How might families react to the app?
  - How much would an app like this get used?
  - What content might the app include?
- Ask the clinician if they have any questions or if there are any points that they would like to raise/discuss before the end of the interview.

FINAL ACTIONS

Reiterate the plan following interview i.e. interviews with other participants, data analysis and the dissemination plan.

"How have you found the interview" – as a transition 'out' of the interview.

Appendix F – HRA/REC favourable opinion letter for qualitative study

Ymchwil Iechyd a Gofal Cymru
Health and Care Research Wales

NHS
Health Research Authority

Professor Anthony Redmond
Professor of Clinical Biomechanics
University of Leeds
Leeds Institute of Theumatic and Musculoskeletal Medicine
School of Medicine, University of Leeds
Level 2, Chapel Allerton Hospital, Leeds
LS7 4SA

Email: approvals@hra.nhs.uk
HCRW.approvals@wales.nhs.uk

02 December 2021

Dear Professor Redmond

HRA and Health and Care Research Wales (HCRW) Approval Letter

Study title:	**What are the experiences of key stakeholders in NON-STOP (Non-Surgical Treatment of Perthes' Disease): A qualitative study**
IRAS project ID:	**300407**
REC reference:	**21/WS/0138**
Sponsor	**University of Leeds**

I am pleased to confirm that **HRA and Health and Care Research Wales (HCRW) Approval** has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications received. You should not expect to receive anything further relating to this application.

Please now work with participating NHS organisations to confirm capacity and capability, in line with the instructions provided in the "Information to support study set up" section towards the end of this letter.

How should I work with participating NHS/HSC organisations in Northern Ireland and Scotland?
HRA and HCRW Approval does not apply to NHS/HSC organisations within Northern Ireland and Scotland.

If you indicated in your IRAS form that you do have participating organisations in either of these devolved administrations, the final document set and the study wide governance report

(including this letter) have been sent to the coordinating centre of each participating nation. The relevant national coordinating function/s will contact you as appropriate.

Please see IRAS Help for information on working with NHS/HSC organisations in Northern Ireland and Scotland.

How should I work with participating non-NHS organisations?
HRA and HCRW Approval does not apply to non-NHS organisations. You should work with your non-NHS organisations to obtain local agreement in accordance with their procedures.

What are my notification responsibilities during the study?

The standard conditions document "*After Ethical Review – guidance for sponsors and investigators*", issued with your REC favourable opinion, gives detailed guidance on reporting expectations for studies, including:

- Registration of research
- Notifying amendments
- Notifying the end of the study

The HRA website also provides guidance on these topics, and is updated in the light of changes in reporting expectations or procedures.

Who should I contact for further information?
Please do not hesitate to contact me for assistance with this application. My contact details are below.

Your IRAS project ID is **300407**. Please quote this on all correspondence.

Yours sincerely,

Natalie Wilson
Approvals Manager

Email: approvals@hra.nhs.uk

Copy to: *Mrs Jean Uniacke, Sponsor contact*

List of Documents

The final document set assessed and approved by HRA and HCRW Approval is listed below.

Document	*Version*	*Date*
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [Uni insurance]		29 September 2021
Interview schedules or topic guides for participants [Topic guide child/family v1]	1	28 September 2021
Interview schedules or topic guides for participants [Topic guide clinician v1]	1	28 September 2021
IRAS Application Form [IRAS_Form_05102021]		05 October 2021
Letter from sponsor [Sponsor agreement]		
Letters of invitation to participant [Invitation for clinicians v1]	1	28 September 2021
Letters of invitation to participant [Email inviting parent v1.1 (tracked)]	1.1	29 November 2021
Letters of invitation to participant [Email inviting parent v1.1]	1.1	29 November 2021
Organisation Information Document [OID]	1	01 October 2021
Participant information sheet (PIS) [PIS clinician v1.1 (tracked)]	1.1	29 November 2021
Participant information sheet (PIS) [PIS parent v1.1 (tracked)]	1.1	29 November 2021
Participant information sheet (PIS) [PIS Clinician v1.1]	1.1	29 November 2021
Participant information sheet (PIS) [PIS Parent]	1.1	29 November 2021
Participant information sheet (PIS) [PIS Child v1.1]	1.1	29 November 2021
Participant information sheet (PIS) [PIS child v1.1 (tracked)]	1.1	29 November 2021
Research protocol or project proposal [Protocol]	1	28 September 2021
Response to Request for Further Information [Responses to REC]		
Schedule of Events or SoECAT [SoECAT]	1	01 October 2021
Summary CV for Chief Investigator (CI) [CI CV - Professor Anthony C Redmond]		14 February 2020
Summary CV for student [Lead researcher CV - Adam Galloway]		
Summary of any applicable exclusions to sponsor insurance (non-NHS sponsors only) [Uni insurance]		01 October 2021

Appendix G – Participant quote table from interview study

Quote table for qualitative study

Code	Theme	Participant quoting
1	**Outcomes**	
1.1	Defining outcomes	
	It tends to be the active, happy children whose parents are engaging and want their child to be better, in my experience, that have the best outcome.	Surgeon, Essex
	I'm thinking about the child, I don't want them to be in pain, I don't want them to be limping, I don't want them to be off school for six months so that they get mental health issues, which we're seeing a lot of right now.	Surgeon, Essex
	I genuinely don't know what's successful and what's not successful	Surgeon, Alder Hey
	Improvement in function which translates into better quality of life for them to play in the playground with their friends, to take part in sports, to be pain free, to have a good night's sleep. I also hope that I'm improving their hip enough to salvage it for further on in their growth and into adulthood	Physio, Harrogate
	he did say if it means getting rid of the pain, I'll have the operation and I'll have a new hip	Grandmother of male, 7yo, Hull
	At every point along the way we've had a choice, haven't we? So with the osteotomy, even though that was what was recommended and there was still a choice	Mother of 16yo, Alder Hey
	I did feel like we could have said, no. We did have an option	Mother of 9yo, Leeds
1.2	Rationale for treatment	
	there's research that says potentially we could offer you…there's some evidence that it might improve things, but in reality… Because that was on the basis of radiologic outcomes, wasn't it, not functional outcomes? So in reality, is that very good evidence for it.	Surgeon, Hull
	I've got to put my hands up and say I do very little with these kids now because over time you know that these kids, a lot of them will come out the other end no matter what you do with their own outcome	Physio, Sheffield
	these kids are in pain, it's limiting their function, it's impacting their…they're getting pain daily, they're limping, they're having to use a stick at university, they can't participate in sports. And actually you give them a hip replacement and they're cracking on like nothing's ever wrong and they love it. I had one boy emailed me from climbing Machu Pichu in Peru for his follow-up PROMs data and he'd had it for Perthes.	Surgeon, Essex
	my approach is very much, very much to, kind of, let your kid be normal.	Surgeon, Alder Hey

2.3	Where the app would get used	
	if you got the odd patient, you might be able to get the information to them via school because they would all be going to school and have access to IT there.	Surgeon, Hull
	we could use them, if need be, in clinics, 'cause I'm sure we'd get charity money for that, for iPads and things like that, if need be. But it would be nice to, if the kid could have it at home.	Physio, Norwich
	it's something you can do independently at home, rather than taking time out of school	CNS, Leeds
	if they're at school and someone doesn't understand, you can just say, well, there's an app, you download the app and you can read everything about it, like, it's there to raise awareness. And I think an app would be a very, very good way to raise awareness because again it's so rare	Mother of 12yo male, Leeds
	And it would help at school, the teachers wouldn't have to do one on one exercises, I can do it in PE, when the other kids are doing activities that I can't do	8yo female, Leeds
3	**Reasons for app use**	
3.1	Previously successful/beneficial apps	
	we do use an iAdjust app for the youngsters that have to do the programme of turns, in Mr Foster's clinic. And that's used, and the kids get involved by doing it by themselves	CNS, Leeds
	[do you think it's better than the old homework you used to get for maths?] yes, because it's fun and it's not like normal maths	8yo female, Leeds
	Spelling Shed is, they give you words, like, they record a word and then, like, someone says it and you have to guess how you spell it and you get points every time. There are leader boards, so you can be, like, first in your class or first in the school	8yo male, Alder Hey
	Times Table Rock Stars, or something. It's quite fun	10yo male, Alder Hey
	I think apps is something that they're more comfortable with, so I think it's a great idea	Father of 16yo male, Alder Hey
	If we could use it in a similar way maybe to how the rheumatology self-management apps work, I think that might improve compliance.	Physio, Harrogate
3.2	Doing something rather than nothing	
	And so giving a patient some kind of intervention so that they feel empowered that they're actually doing something to manage their own treatment	Surgeon, Hull

	the older presentation ones, the ones that are your eight plus, nine, and I just think sometimes no matter what you do, movement exercises, it doesn't improve things.	Physio, Hull
	The financial ability of the parent and their time to be able to take the kid swimming, to access swimming, to access cycles – they may not be able to afford a bike.	Physio, Harrogate
2	**Who, when, where**	
2.1	Different users of the app	
	Most children have their own phones or appear to that come here, so whether it was on theirs or their parents' or both	Physio, Hull
	actually most three- and four-year-olds can navigate a smartphone, which is as depressing as hell	Physio, Fife
	that would be useful for the children because they're used to using apps and it would be useful for the parents because it would enable them to try and take a bit of ownership of the condition	Surgeon, Bristol
	an app might be a useful way for parents to engage and to feel that someone cares about them	Surgeon, Alder Hey
	The other thing I love with apps is that it enables the child to take some ownership and some responsibility and they have therefore some understanding of what they're trying to achieve. No matter how young the kids are, they want to be involved in their own care on the whole, I've found. And I think that's important to acknowledge that and to respect that and to enable that. So yeah, bloody love an app	Physio, Harrogate
	I know you're going to do an app that's going to be for the children, but even for parents too because there's a lot of scaremongering goes on	Mother of 16yo female, Alder Hey
2.2	When the app gets used	
	I think it would get used a lot initially and I think it will tail off, just because they then do well or better hopefully and the same with any physio	Physio, Hull
	it's encouraging them to be more autonomous in their care, because that's teenage transition as well, We're teaching young people to take responsibility for their actions	CNS, Leeds
	I think it would be used different amounts at different stage of disease	Surgeon, Alder Hey
	when we got given the physio initially, it was really time-consuming for me to do the physio so if the child had their own app to follow, doing it themselves, it just frees up your parent's time	Mother of 12yo male, Leeds

	I think that sometimes you get a diagnosis of Perthes, and if your orthopaedic surgeon is going down a nonsurgical route, I think that in one way the parents and the children are pleased they don't need the surgery, but in another way then they're not having, in their mind, an active treatment	Surgeon, Bristol
	I think an app might be a useful way for parents to engage and to feel that someone cares about them	Surgeon, Alder Hey
	it would be nice if families could, could have some exercises. Could have some ways of feeling that they were empowered	Surgeon, Alder Hey
3.3	Rewards and levels	
	having something in it that incentivises doing the exercises. If your app is going to be for the child, making it colourful and exciting so they win something. I don't know, give them Roblox money or something! If you do your exercises, you get something for Fortnite, things like that.	Surgeon, Hull
	So if they're doing their exercises and mum ticks it, or they've ticked it, to say, yeah, I've done them, you're gathering rewards or tokens, free books. I think reward-driven, it's got to give them something at the end of it, hasn't it?	Physio, Sheffield
	I think good interaction, fun, animated characters, that kids can relate to. So it might have to be updated as characters come in from…you know, like, it's Paw Patrol at the moment,	CNS, Leeds
	Rewards, yeah, and you could get points every time and then you can use those points to make your Avatar or design, like, your house, or something and also it, like, reminds you to do exercise every day and do your daily exercise	8yo male, Alder Hey
	I don't know if he'd prefer to design it himself or choose, like, a Roblox character or something to invent himself.	Mother of 11yo male, Hull
	You like getting your stickers when you go to physio, don't you, you could maybe have some virtual stickers or something like that	Mother of 6yo male, Leeds
4	**Core features of an app**	
4.1	Educational resources regarding condition	
	I would imagine that there would be a degree of education, and that would be an appropriate thing both for kids and for adults, and information is always good	Surgeon, Bristol
	I think that'd be good, yes, if there was more information in one place [an app]. One that isn't just like a scary site of mums all freaking out	Mother of 9yo female, Leeds

	if they're at school and someone doesn't understand, you can just say, well, there's an app, you download the app and you can read everything about it, like, it's there to raise awareness. And I think an app would be a very, very good way to raise awareness because again it's so rare	Mother of 12yo male, Leeds
	he can't go and join a football team, he's not allowed to go on a bouncy castle, if he could have a bit more understanding of why he can't do those things, I don't know if that would help, but that's probably the one thing that he really, really, struggles with	Mother of 6yo male, Leeds
	I don't know if it needs to be…from a parents' point of view if it needs to be massively medical and technical because you can wind yourself up into something	Mother of a 12yo male, Hull
4.2	Communication	
	Most orthopods don't take direct communication from parents and families, it's just too much	Surgeon, Bristol
	it would be nice if families had a way to connect with each other to compare notes and discuss what's going on. I think it would be nice if families could somehow, keep a diary of what was going on, so, they could show their consultant	Surgeon, Alder Hey
	it would be really nice if there was a way of pushing that data towards their consultant or, kind of, communicating with someone that cares	Surgeon, Alder Hey
	They could have little pen-friends with each other or a little chat room with each other, just, I'm feeling rubbish today because I didn't sleep last night or I'm feeling rubbish because I've watched my friends at PE today at school, do you know, just Perthes' to Perthes'	Mother of 9yo female, Leeds
	Like a little whinging room for mums so they can moan at each other and a whinging room for kids on how rubbish is it not sleeping or not playing sports or how annoying is my mum telling me no all the time?	Mother of 9yo female, Leeds
4.3	Self-management within an app	
	giving a patient some kind of intervention so that they feel empowered that they're actually doing something to manage their own treatment or their child's treatment I think will go down really well. I think there will be a proportion of people who aren't bothered, but I think that would be maybe less than a quarter would be those kinds of patients.	Surgeon, Hull
	But do I want an app telling me what I should do for my patients? Hell no.	Surgeon, Essex

	The other thing I love with apps is that it enables the child to take some ownership and some responsibility and they have therefore some understanding of what they're trying to achieve. No matter how young the kids are, they want to be involved in their own care on the whole, I've found. And I think that's important to acknowledge that and to respect that and to enable that. So yeah, bloody love an app.	Physio, Harrogate
	it's encouraging them to be more autonomous in their care, because that's teenage transition as well, isn't it? We're teaching young people to take responsibility for their actions	CNS, Leeds
	If I get in pain or whatever, I would probably go onto the app and do all the things on it.	12yo male, Leeds
	when we got given the physio initially, it was really time-consuming for me to do the physio so if the child had their own app to follow, doing it themselves, it just frees up your parent's time	Mother of 8yo female, Leeds
5	**Variation of care**	
5.1	Current usual care	
	my mainstay of treatment is to maintain their range of motion, make sure that their pain is controlled, and to let them have as normal life as possible.	Surgeon, Hull
	if they've got a good range of abduction, I don't routinely refer them to physio. If they've got a decreased range of abduction then I refer them to physio and ask the physio to assess them for hydro if they have that available to them.	Surgeon, Bristol
	[What do you like about the treatment you do for your hip?] I get to swim more	9yo female, Leeds
	early physiotherapy I feel is really important.	Surgeon, Essex
	our approach is to avoid bouncy castles and trampolines, but otherwise let them have a normal, a normal life.	Surgeon, Alder Hey
	Do whatever you want but restricting bouncy castles and trampolines. There's no evidence for it but it, kind of, seems, kind of, sensible and they're easy to avoid.	Surgeon, Alder Hey
	Aqua works well, because it's easy for them, it's lovely, you can have fun	Physio, Kent
5.2	Different approaches	
	In the early years in my practice, when I was quite keen, I probably operated on more than I would now. And I have a suspicion that they're the ones that had the good outcomes, so they're probably the ones that if I left alone would probably have done quite well as well.	Surgeon, Hull

	as orthopaedic surgeons our whole career we've been told, no, no, no, you can't do an early hip replacement, you have to wait until they can't walk anymore and in terrible pain. But actually younger people do very well and it gives them a new lease of life, so it's not the worst outcome in the world if that's what they end up having	Surgeon, Hull
	practice is changing. It used to be, we talked about slings and springs years and years ago, trying to keep range of motion going and then, I don't think slings and springs was probably taught in physiotherapy colleges for some years. And then, lo and behold, my consultant were saying, oh, any chance of doing something with slings and springs? So like it's gone a full circle.	Physio, Norwich
	we stopped doing the broomstick casting, so that was really helpful	Physio, Fife
	When I first started work all the information was no impact	Physio, Harrogate
	We tried doing a class at one point. We did a Perthes' class. We had about 20 kids at one point, it was ridiculous	Physio, Sheffield
5.3	Evidence to support decision making	
	I think ruining some poor child's childhood with lots of big femoral osteotomy when we don't necessarily know that they really make a difference to their function or the age that they're going to have a hip replacement, it doesn't seem like a very good thing to be doing to them.	Surgeon, Hull
	you can't manage anything unless you have information	Physio, Fife
	you see a child with Perthes and you genuinely don't really know in your heart of hearts the best treatment algorithm for them	Surgeon, Bristol
	I'd like to know whether surgery made a difference or not	Surgeon, Alder Hey
	even if it didn't make a difference to the hip, so, to the shape of the hip which is what surgeons care about, I'd be interested to see whether surgery made the difference to patient outcomes. To, kind of, pain and stuff. Because I don't know that.	Surgeon, Alder Hey
	if you know how long you're going to have it for you will be like me, I'm just…I don't know, I'm just waiting for it to go, if you know when it's going to go probably, you're waiting for your best day of your life basically.	10yo male, Alder Hey
5.4	Agreement amongst clinicians	
	it would be really nice if we could move towards some consistency or consensus of how the patient should be managed so that we're all giving the same kind of information.	Surgeon, Hull

	I'd like to see is some consistency so that a) we've got some guidance as to, look, this is what you should be doing, and then have a large proportion of people doing it for all patients	Surgeon, Hull
	I certainly didn't agree with the [other treating centre's] ethos of let's put them in a wheelchair for a year, because I don't think that made any difference to the outcome	Surgeon, Hull
	there are so many different treatments, nobody agrees	Surgeon, Hull
	I think BSCOS obviously should and will want to be involved in this. They'll probably just set up another Delphi consensus group, to be perfectly honest, which will take three years to sort out. Because especially with Perthes, because it's probably one of the most varied treatment managements that we see	Surgeon, Essex
	that's what [my consultant] said. He was like, I'm so sorry, if you go and see any consultant, we'll all say something different	Mother of 10yo male, Alder Hey
6	**AOB**	
6.1	COVID impact	
	We used to use hydrotherapy but we unfortunately don't have a pool anymore, it was closed during COVID and it's not looking like it's going to open	Physio, Hull
	silver lining of COVID is everybody's become so much more au fait with technology	Physio, Fife
	if I make a decision that I want them to have an arthrogram that's a time dependant decision for me, so I'll list them to be done within three months, and generally I can. Obviously, there's pressure on services at the moment and waiting lists	Surgeon, Bristol
	we've had COVID for two years so we, kind of, haven't been seen. We've just been shoved on a shelf	Mother of 9yo female, Leeds
	I do think for us lockdown became quite a positive thing for Perthes' because like I say, he just couldn't…I think that's what the best thing was, was just to rest it	Mother of 12yo male, Hull

Appendix H – Thematic table for interview study

This table shows a frequency count for each participant within the interview study and shows the number of responses each participant gave in each theme/subtheme.

Thematic table for clinician participants

Code \ Participant	Clin1	Clin2	Clin3	Clin4	Clin5	Clin6	Clin7	Clin8	Clin9	Clin10	Clin11	Clin12
1.1 Defining outcomes	3	4	7	4		2		2	1			2
1.2 Rationale for treatment	4	7	4	1	1	2		1	2	1	1	2
2.1 Different users of the app	1	2		3	1	3	3	3	3		2	1
2.2 When the app gets used	1			2	1	1	1	2	1		2	
2.3 Where the app would get used	2			1		1					2	1
3.1 Previously successful/beneficial apps	1				1				3		2	
3.2 Doing something rather than nothing	1		1				2	2				
3.3 Rewards and levels	2	4	2	4	2	1	1		2	2	5	
4.1 Educational resources regarding condition	4	1	4	1	2	1	2	3		2		2
4.2 Communication with parties involved (peer or clinical)		2	3	4	1	1	3	2	1		1	
4.3 Self-management within an app	1	1	2	5	3	2	2	1	1	4	2	2
5.1 Usual care currently		5	11	2	2	1	3	8	4	1	2	3
5.2 Different approaches used or experienced	6	10	6	3	1	3		1	2	5	1	
5.3 Evidence to support decision making	6	2	3	3	4	1	3	3	4	2		1
5.4 Agreement amongst clinicians	2	4	4	1	1		2		1			
6.1 COVID impact	1			1	1	1	1					

Thematic table for child/family participants

Code \ Participant	Child1	Child2	Child3	Child4	Child5	Child6	Child7	Child8	Child9	Child10	Child11	Child12
1.1 Defining outcomes	1	3		2	1	1	2		1			
1.2 Rationale for treatment			1	1		1						
2.1 Different users of the app										1	2	1
2.2 When the app gets used									1		1	
2.3 Where the app would get used				1							1	
3.1 Previously successful/beneficial apps	3		2	1	2	2	1	1	1		3	1
3.2 Doing something rather than nothing												
3.3 Rewards and levels	1		3	4	4	1		2	2			2
4.1 Educational resources regarding condition	1	1	1	1		1	5	3	3	1	1	4
4.2 Communication with parties involved (peer or clinical)	2	3		2			1		2			1
4.3 Self-management within an app	1	1	2	3	3	3	1	1	2	2	2	5
5.1 Usual care currently							1					
5.2 Different approaches used or experienced	1			2		1	2					
5.3 Evidence to support decision making		1		1		1	2		2			1
5.4 Agreement amongst clinicians	2						1			1		
6.1 COVID impact	1											2

Appendix I – Logic models throughout project

Preliminary logic model — NON-STOP Logic model v1

Problem addressed

Based on current evidence, there is a need for the development of a digital self-management intervention for children with Perthes' Disease.

Priorities for intervention

1. *Previous case review highlighting significant variation of 'usual' non-surgical management for Perthes' Disease in the UK* [1]
2. *Based on systematic review that found no robust evidence to support the use of any non-surgical intervention compared with another* [2]
3. *Digital intervention ('app') designed using evidence-based guidance (MRC, PPI, theoretical input)* [3]
4. *James Lind Alliance identified need for research exploring outcomes following non-surgical care of Perthes' Disease* [4]

Inputs	Outputs (activity)	Change mechanisms	Measuring change	Impact
Children/families - Participants following self-management support - Videos/examples of exercises to be used - Involved in the JLA priority setting for this topic - Access to smart phones Clinicians - Children/families regularly reviewed in outpatients - Input to identify important content for app - Training staff on app use - Clinicians' training children/families to use app Intervention (the app) - App developers' expertise on 'what works' in app design	App design - Early versions of app, including training for app, led by developers. - Clinical content of app informed by an umbrella review & clinical consensus work (Delphi study) - App design tested iteratively Early testing of app - Clinicians trained on how to use/support children/families - Children/families test out app (acceptability, feasibility of use)	- Umbrella review & consensus work will identify change mechanisms supporting to maximise engagement with app - Self-determination theory: Changes in 'psychological needs' – increasing the motivation of the child/family to complete their physiotherapy - Exercises will strengthen and lengthen relevant muscles and in turn, stabilise hip joint	Process measures - Metrics from app measuring usage (how many times used, how long for, what has been accessed when logged on). - Qualitative feedback on use of app from children/families and clinicians Outcome measures - PROMs from Core Outcome Set - Radiological outcomes (Stulberg or equivalent)	- App that can promote improve self-management behaviour - Improved clinical and PROM outcomes - Potential to reduce need for surgical intervention - A plan for a definitive clinical trial

Contextual factors

Resources
- Remote self-management increasingly common in rehab settings post-COVID.
- Time for children/families to do the exercises/aspects of the app.
- Time for clinicians to train children/families on how to use the app.
- Access to internet/smartphone to use the app
- Could app be used outside of family settings e.g., in schools, or other activities?

Service
- Some centres don't have specialist clinics where these patients are 'located' clinically. So could lose out due to difficult making clinicians aware of intervention.

1. Galloway, A.M., et al., *A case review to describe variation in care following diagnosis of Perthes' disease.* 2020. **1**(11): p. 691-695
2. Galloway, A.M., et al., *A systematic review of the non-surgical treatment of Perthes' disease.* 2020. **1**(12): p. 720-730.
3. Skivington K, Matthews L, Simpson S A, Craig P, Baird J, Blazeby J M et al. A new framework for developing and evaluating complex interventions: update of Medical Research Council guidance BMJ 2021; 374: n2061 doi:10.1136/bmj. n2061
4. Vella-Baldacchino, M., et al., *Research priorities in children requiring elective surgery for conditions affecting the lower limbs: a James Lind Alliance Priority Setting Partnership.* BMJ Open, 2019. 9(12): p. e033233

Interim logic model NON-STOP Logic model v2

Problem addressed

Based on current evidence, there is a need for the development of a digital self-management intervention for children with Perthes' Disease.

Priorities for intervention

1. *Previous case review highlighting significant variation of 'usual' non-surgical management for Perthes' Disease in the UK* [1]
2. *Based on systematic review that found no robust evidence to support the use of any non-surgical intervention compared with another* [2]
3. *Digital intervention ('app') designed using evidence-based guidance (MRC, PPI, theoretical input)* [3], *qualitative interview study* [5]
4. *James Lind Alliance identified need for research exploring outcomes following non-surgical care of Perthes' Disease* [4]

Inputs	Outputs (activity)	Change mechanisms	Measuring change	Impact
Children/families - Participants following self-management support - Videos/examples of exercises to be used - Involved in the JLA priority setting for this topic - Access to smart phones - Qualitative work highlighted importance of inclusion in decision making Clinicians - Children/families regularly reviewed in outpatients - Input to identify important content for app - Training staff on app use - Clinicians' training children/families to use app - Empowering patients important to clinicians (from qualitative work) Intervention (the app) - App developers' expertise on 'what works' in app design - Empowerment and patient control important (from qualitative work)	App design - Early versions of app, including training for app, led by developers. - Clinical content of app informed by an umbrella review & clinical consensus work (Delphi study) - App design tested iteratively Early testing of app - Clinicians trained on how to use/support children/families - Children/families test out app (acceptability, feasibility of use) - Look to utilise PPI groups (PAG & YPAG) to do some informal testing of app – usability and acceptability	- Consensus work will identify change mechanisms supporting to maximise engagement with app - Self-determination theory: Changes in 'psychological needs' – increasing the motivation of the child/family to complete their physiotherapy - Exercises will strengthen and lengthen relevant muscles and in turn, stabilise hip joint	Process measures - Metrics from app measuring usage (how many times used, how long for, what has been accessed when logged on). - Qualitative feedback on use of app from children/families and clinicians Outcome measures - PROMs from Core Outcome Set (PROMIS mobility) - Radiological outcomes (Stulberg or equivalent) - Clinical markers such as ROM and strength. Pain captured in PROM	- App that can promote improve self-management behaviour - Improved clinical and PROM outcomes - Potential to reduce need for surgical intervention - A plan for a definitive clinical trial

Contextual factors

Resources
- Remote self-management increasingly common in rehab settings post-COVID.
- Time for children/families to do the exercises/aspects of the app.
- Time for clinicians to train children/families on how to use the app.
- Access to internet/smartphone to use the app
- Could app be used outside of family settings e.g., in schools, or other activities?

Service
- Some centres don't have specialist clinics where these patients are 'located' clinically. So could lose out due to difficult making clinicians aware of intervention. Although qualitative work did not highlight any obvious issues

1. Galloway, A.M., et al., *A case review to describe variation in care following diagnosis of Perthes' disease.* BJO. 2020. **1**(11): p. 691-695
2. Galloway, A.M., et al., *A systematic review of the non-surgical treatment of Perthes' disease.* BJO. 2020. **1**(12): p. 720-730.
3. Skivington K, Matthews L, Simpson S A, Craig P, Baird J, Blazeby J M et al. A new framework for developing and evaluating complex interventions: update of Medical Research Council guidance. BMJ. 2021; 374: n2061 doi:10.1136/bmj. n2061
4. Vella-Baldacchino, M., et al., *Research priorities in children requiring elective surgery for conditions affecting the lower limbs: a James Lind Alliance Priority Setting Partnership.* BMJ Open, 2019. 9(12): p. e033233
5. Galloway, A.M., et al., *"Waiting for the best day of your life". A qualitative interview study of patients' and clinicians' experiences of Perthes' disease.* Bone & Joint Open, 2023. **4**(10): p. 735-741.

Appendix J – Summary of evidence for participants

Summary of evidence for Non-Surgical Treatment of Perthes' (NON-STOP) Delphi study

Thank you for considering taking part in the Delphi study to achieve clinical consensus on NON-STOP. This document provides a brief summary of the available evidence that is relevant to this study as well as links to full texts where possible. The aim is to provide some clinical context prior to the start of the survey that will follow, which includes statements around NON-STOP for you to vote on, displaying your level of agreement/disagreement. The domains within the survey have been designed based on the evidence provided, as well as input from key stakeholders including specialist clinicians and patient/public involvement.

1. *Qualitative study of key stakeholders (unpublished work)*

This study is in now complete and in the write-up stage. It involved interviews with clinicians and children with Perthes' Disease and their families, the questions aimed to explore their experiences of NON-STOP as well as their thoughts on what future care should look like including ideas on a digital self-management intervention (an app). A summary of the responses that arose are:

- Clinicians and child/family dyads need consistent advice based on evidence.
- Long and short-term goals were discussed i.e., treatment that impacts both radiological outcome at skeletal maturity and function i.e., pain, activity levels. With the focus from both being quality of life in the first instance.
- The idea of an app well received as a concept by clinicians and child/family dyads.
- An app could provide a step towards consensus/agreement on treatment and a reduction in variation of care.

2. *Systematic review of NON-STOP (https://tinyurl.com/NON-STOPSR)*

This systematic review compared the effectiveness of non-surgical interventions against one another. It looked at a range of interventions including active observation, physiotherapy, bracing/casting and activity modification/weightbearing change. There was no robust evidence to support one NON-STOP compared to another with majority of studies having significant issues with methodological quality and bias. Some studies showed improvement in domains like range of motion and strength with physiotherapy input, however it did not correlate with radiological changes at skeletal maturity and outcomes such as quality of life and function were not measured. Overall the paper concludes that more research in NON-STOP is needed.

3. *Case review: variation of care in Perthes' Disease (https://tinyurl.com/NON-STOPCR)*

A case review looking at five centres in the UK demonstrated widespread variation of care in the UK when assessing things such as advice on pain relief and activity modification, input from physiotherapy locally or regionally and how often they are seen by orthopaedic specialists (physiotherapy and medical).

4. *The BOSS Study results (https://tinyurl.com/BOSSresults)*

The British Orthopaedic Surgery Surveillance study provided data on incidence and rate of surgical intervention in all but one hospital in the UK. It showed that despite a third of patients receiving surgery, there were no evidence of improved outcomes in quality of life or Stulberg. It did however demonstrate appropriate numbers for a future randomised trial, which this Delphi study would inform for the conservative management arm.

5. *Herring, 2004 (https://pubmed.ncbi.nlm.nih.gov/15466720/)*

One of only two prospective cohort studies, studied surgery and no surgery and found that children aged >8 years old did better with surgery however females of any age, and those aged >8 years old did worse than those younger, irrespective of gender or treatment type. Also demonstrated no significant difference in outcomes when looking at NON-STOP.

6. *Wiig, 2008 (https://pubmed.ncbi.nlm.nih.gov/18827249/)*

This prospective study suggested that children over the age of six years old at diagnosis with >50% femoral head involvement had better outcomes from surgery compared to physiotherapy or orthosis. As with the Herring study, this study relied on post-hoc analyses and therefore are at risk of type I error.

7. *Perthes' core outcome set (https://pubmed.ncbi.nlm.nih.gov/32349599/)*

This piece developed a set of outcomes to be employed in future studies in Perthes' Disease. They did so by carrying out a Delphi study with key stakeholders nationally and internationally and finished with 16 outcomes derived from 6 categories (life impact, resource use, pathophysiological manifestations, death and technical considerations). They also provided the PROMIS mobility as a valid tool in this population.

Appendix K – Initial survey for clinical consensus study

1. **Exercises.** ***The following statements will be based on exercise advice/instruction that children with Perthes' Disease should be encouraged to adhere to.***
    a. Strengthening exercises. "Children with "early stage" Perthes' Disease should complete":
        i. hip strengthening exercises
        ii. knee strengthening exercises
        iii. foot & ankle strengthening exercises
        iv. trunk strengthening exercises
        v. high impact strengthening exercises (e.g. jump-squats, star-jumps)
        vi. Any strengthening exercises as long as they avoid discomfort
    b. Range of movement stretching exercises "Children with "early stage" Perthes' Disease should complete":
        i. hip stretches
        ii. knee stretches
        iii. foot & ankle stretches
        iv. spinal stretches
        v. Any strengthening exercises as long as they avoid discomfort
    c. Strengthening exercises. "Children with "late stage" Perthes' Disease should complete":
        i. hip strengthening exercises
        ii. knee strengthening exercises
        iii. foot & ankle strengthening exercises
        iv. trunk strengthening exercises
        v. high impact strengthening exercises (e.g. jump-squats, star-jumps)
        vi. Any strengthening exercises as long as they avoid discomfort
    d. Range of movement stretching exercises "Children with "late stage" Perthes' Disease should complete":
        i. hip stretches
        ii. knee stretches
        iii. foot & ankle stretches
        iv. spinal stretches
        v. Any strengthening exercises as long as they avoid discomfort
    e. Water-based exercise "Children with Perthes' Disease should be advised to complete":
        i. supervised (physiotherapy-lead) water-based exercises
        ii. water-based exercise as self-management i.e. doing prescribed exercises in a local pool (not supervised by physiotherapist)
        iii. water-based exercise when land-based physiotherapy is not effective
    f. Functional ability exercises "Children with Perthes' Disease should":
        i. complete balance exercises
        ii. receive gait education
        iii. have advice on potential use of mobility aids
    g. Who, when, where? "Children with Perthes' Disease should complete prescribed exercise regimes":
        i. at home under the supervision of parent/family members
        ii. at school under the supervision of school-staff members
        iii. at the hospital under the supervision of clinicians

iv. at extracurricular activities (e.g., sports clubs, etc.) under the supervision of those leading the activities

h. Please enter any additional exercise recommendations that you think are important below.

2. **Physical Activity.** ***The following statements will be based on physical activity advice/instruction that children with Perthes' Disease should be encouraged to adhere to.***
    a. Recreational activities "in the early stages of Perthes' Disease, the following activities should be discouraged":
        i. Swimming
        ii. Contact sports (e.g. football, rugby)
        iii. Other team sports (e.g. basketball, netball, hockey)
        iv. Long distance running (more than 1-2 mile)
        v. Horse riding
        vi. Cycling
        vii. Gymnastics
        viii. PE at school
        ix. High-impact activities (e.g. bouncy castles and trampolines)
    b. Recreational activities "in the later stages of Perthes' Disease, the following activities should be discouraged":
        i. Swimming
        ii. Contact sports (e.g. football, rugby)
        iii. Long distance running (more than 1-2 mile)
        iv. Horse riding
        v. Cycling
        vi. Gymnastics
        vii. PE at school
        viii. High-impact activities (e.g. bouncy castles and trampolines)
    c. Activity modification. "Children with Perthes' Disease should":
        i. use a walking aid (e.g. crutches, Zimmer—frame) to modify their activity if symptoms (pain, limping, reduced activity) persist
        ii. use a wheelchair to modify their activity if symptoms (pain, limping, reduced activity) persist
    d. Please enter any additional physical activity recommendations that you think are important below.

3. **Education/information sharing.** ***This section relates to information that clinicians may deliver to children with Perthes' Disease and their families that falls under the remit of non-surgical treatment. It covers a breadth of the multi-disciplinary team.***
    a. Understanding of Perthes' Disease. "Clinicians should provide children/families with":
        i. information regarding the disease process including the affected anatomical structures and prognosis
        ii. information regarding current research relating to Perthes' Disease including aetiology and epidemiology

iii. information regarding where additional patient and family information resources can be found (e.g. STEPS website)

b. Pain management. "Children with Perthes' Disease should":
   i. be advised to take paracetamol or equivalent for pain management
   ii. be advised to take ibuprofen or equivalent for pain management
   iii. be advised to take morphine or equivalent for pain management
   iv. receive advice on pacing and activity levels
   v. be advised on the use of heat/cold therapy
   vi. be advised on the use of TENS/equivalent electrotherapy
   vii. be encouraged to use massage to aid pain relief
   viii. be prescribed steroid injection for pain management
   ix. be referred to a pain management service if their symptoms are not managed with medication and/or physiotherapy
c. Weight management and nutrition in Perthes' Disease
   i. All children should receive advice on lifestyle, weight management and nutrition from a healthcare professional.
   ii. Children should receive advice on lifestyle, weight management and nutrition only when indicated
   iii. Children should be referred to a specialist service for weight management and nutritional advice when clinically indicated
   iv. Monitoring weight management and nutrition is an essential part of treatment i.e. height and weight at all appointments
d. Mental wellbeing in Perthes' Disease
   i. Children and their families should be given the opportunity to discuss their (or their child's) mental wellbeing with any healthcare professional
   ii. All children should be signposted to general mental wellbeing resources e.g. the STEPS Charity website or NHS 111 website.
e. Please enter any additional education/information recommendations that you think are important below.

4. **Input from other services.** ***This section refers to the input from NHS and non-NHS professionals for children with Perthes' Disease including what should be a part of their treatment and who might provide it.***
   a. Referral to orthopaedics and ongoing management in Perthes' Disease
      i. Any child with suspected Perthes' disease should be referred for specialist review
      ii. Any child who does not improve from a symptom/symptom-management perspective should have access to an orthopaedic specialist
      iii. Children with Perthes' Disease should be centralised to a team with a specialist interest in Perthes' Disease in their geographical region
   b. Referral to physiotherapy and ongoing management of Perthes' Disease. "Children with Perthes' Disease should";
      i. be offered an initial assessment with a physiotherapist
      ii. be seen by a physiotherapist regularly until the disease process is complete/healing is observed

iii. keep regular activity diaries to share with healthcare professionals
iv. be seen by a physiotherapist until they can self-manage independently

c. Multi-disciplinary team input. “Children with Perthes’ Disease should”:
   i. be offered an assessment from an occupational therapist
   ii. be offered an assessment from a social worker
   iii. be offered an assessment from a psychologist
   iv. have a named clinical ‘key worker’ regardless of MDT role
d. Communication between children/families and clinicians. “Children with Perthes’ Disease and their families should”:
   i. have a means of direct communications with clinicians between appointments
   ii. be directed towards means of contacting other children with Perthes’ Disease and their families i.e. peer-support groups/forums
e. School support. “Children with Perthes’ Disease should have access to a”:
   i. physiotherapist in a school setting
   ii. nurse in a school setting
   iii. named school-support staff member
f. Please enter any additional hospital/service-related recommendations that you think are important below.

5. Monitoring assessments for clinical practice with children with Perthes’ Disease.
This section relates to the monitoring that clinicians should carry out when assessing and treating children with Perthes’ Disease.

a. Clinical assessments. “Children with Perthes’ Disease should have”:
   i. their ROM documented at every appointment (regardless of MDT-role)
   ii. a validated quality-of-life assessment tool completed at initial assessment and regular intervals
   iii. regular reviews with an orthopaedic specialist until they reach skeletal maturity
b. Please enter any additional monitoring assessment recommendations that you think are important below.

Appendix L – Summary of results from round 1

NON-STOP Delphi Round one results

In round one of the NON-STOP Delphi study there were 41 participants who responded to the survey. They consisted of:

22 Physiotherapists (53.7%)
18 Orthopaedic surgeons (43.9%)
1 Clinical nurse specialist (2.4%)

In the first round, there were 87 statements that were put to the cohort for consideration. 'Clinical consensus' was determined as any statement that achieved more than 75% agreement/disagreement and this was achieved in 31 statements after round one. The statements that achieved consensus will be removed from round two. There were an additional three statements that were included as a result of the free-text comments from round one. It is worth pointing out that there were other comments made, but these were deemed to be outside of the scope of this Delphi study (i.e. MRI scanning, blood tests, etc.).

Here are the domains and statements that achieved consensus after the first round of the survey.

9 in 'Exercises'

Statement	Consensus (agree/disagree)
Children with 'early stage' Perthes' Disease should complete high impact strengthening exercises (e.g. jump-squats, star-jumps)	Disagree
Children with 'early stage' Perthes' Disease should complete hip stretches	Agree
Children with 'late stage' Perthes' Disease should complete hip strengthening exercises	Agree
Children with 'late stage' Perthes' Disease should complete trunk strengthening exercises	Agree
Children with 'late stage' Perthes' Disease should complete hip stretches	Agree
Children with Perthes' Disease should complete water-based exercise as self-management i.e. doing prescribed exercises in a local pool (not supervised by physiotherapist)	Agree
Children with Perthes' Disease should complete water-based exercise when land-based physiotherapy is not effective	Agree
Children with Perthes' Disease should have advice on potential use of mobility aids	Agree
Children with Perthes' Disease complete prescribed exercise regimes at home under the supervision of parent/family members	Agree

5 in 'Physical activity'

Statement	**Consensus (agree/disagree)**
In the early stages of Perthes' Disease, swimming should be discouraged	Disagree
In the early stages of Perthes' Disease, long-distance running (more than 1-2 miles) should be discouraged	Agree
In the later stages of Perthes' Disease, swimming should be discouraged	Disagree
In the later stages of Perthes' Disease, cycling should be discouraged	Disagree
Children with Perthes' Disease should use a walking aid (e.g. crutches, Zimmer—frame) to modify their activity if symptoms (pain, limping, reduced activity) persist	Agree

10 in 'Education/information sharing'

Statement	**Consensus (agree/disagree)**
Clinicians should provide children/families with information regarding the disease process including the affected anatomical structures and prognosis	Agree
Clinicians should provide children/families with information regarding current research relating to Perthes' Disease including aetiology and epidemiology	Agree
Clinicians should provide children/families with information regarding where additional patient and family information resources can be found (e.g. STEPS website)	Agree
Children with Perthes' Disease should be advised to take paracetamol or equivalent for pain management	Agree
Children with Perthes' Disease should be advised to take morphine or equivalent for pain management	Disagree
Children with Perthes' Disease should receive advice on pacing and activity levels	Agree
Children with Perthes' Disease should be advised on the use of heat/cold therapy	Agree
Children with Perthes' Disease should be referred to a specialist service for weight management and nutritional advice when clinically indicated	Agree
Children with Perthes' Disease and their families should be given the opportunity to discuss their (or their child's) mental wellbeing with any healthcare professional	Agree
All children with Perthes' Disease should be signposted to general mental wellbeing resources e.g. the STEPS Charity website or NHS 111 website.	Agree

5 in 'Input from other services'

Statement	**Consensus (agree/disagree)**
Any child with suspected Perthes' disease should be referred for specialist review	Agree
Any child who does not improve from a symptom/symptom-management perspective should have access to an orthopaedic specialist	Agree
Children with Perthes' Disease should be offered an initial assessment with a physiotherapist	Agree
Children with Perthes' Disease and their families should have a means of direct communications with clinicians between appointments	Agree
Children with Perthes' Disease and their families should be directed towards means of contacting other children with Perthes' Disease and their families i.e. peer-support groups/forums	Agree

2 in 'Monitoring assessments'

Statement	**Consensus (agree/disagree)**
Children with Perthes' Disease should have their ROM documented at every appointment (regardless of MDT-role)	Agree
Children with Perthes' Disease should have a validated quality-of-life assessment tool completed at initial assessment and regular intervals	Agree

Appendix M – Clinical consensus recommendations summary graphic

Appendix N – GUIDED checklist for NON-STOP development

GUIDED – a guideline for reporting for intervention development studies.

Supplementary File 1: Blank Checklist

Item description	Explanation	Page in manuscript where item is located	Other*
1. Report the context for which the intervention was developed.	Understanding the context in which an intervention was developed informs readers about the suitability and transferability of the intervention to the context in which they are considering evaluating, adapting or using the intervention. Context here can include place, organisational and wider socio-political factors that may influence the development and/or delivery of the intervention (15).	Chapters 1, 2 & 5	
2. Report the purpose of the intervention development process.	Clearly describing the purpose of the intervention specifies what it sets out to achieve. The purpose may be informed by research priorities, for example those identified in systematic reviews, evidence gaps set out in practice guidance such as The National Institute for Health and Care Excellence or specific prioritisation exercises such as those undertaken with patients and practitioners through the James Lind Alliance.	Chapter 5	
3. Report the target population for the intervention development process.	The target population is the population that will potentially benefit from the intervention – this may include patients, clinicians, and/or members of the public. If the target population is clearly described then readers will be able to understand the relevance of the intervention to their own research or practice. Health inequalities, gender and ethnicity are features of the target population that may be relevant to intervention development processes.	Chapters 1 & 5	
4. Report how any published intervention development approach contributed to the development process	Many formal intervention development approaches exist and are used to guide the intervention development process (e.g. 6Squid (16) or The Person Based Approach to Intervention Development (17)). Where a formal intervention development approach is used, it is helpful to describe the process that was followed, including any deviations. More general approaches to intervention development also exist and have been categorised as follows (3):- Target Population-centred intervention development; evidence and theory-based intervention development; partnership intervention development; implementation-based intervention development; efficacy-based intervention development; step or phased-based intervention development; and intervention-specific intervention development (3). These approaches do not always have specific guidance that describe their use. Nevertheless, it is helpful to give a rich description of how any published approach was operationalised	Chapters 2 & 5	
5. Report how evidence from different sources informed the intervention development process.	Intervention development is often based on published evidence and/or primary data that has been collected to inform the intervention development process. It is useful to describe and reference all forms of evidence and data that have informed the development of the intervention because evidence bases can change rapidly, and to explain the manner in which the evidence and/or data was used. Understanding what evidence was and was not available at the time of intervention development can help readers to assess transferability to their current situation.	Chapters 1, 2 & 5	
6. Report how/if published theory informed the intervention development process.	Reporting whether and how theory informed the intervention development process aids the reader's understanding of the theoretical rationale that underpins the intervention. Though not mentioned in the e-Delphi or consensus meeting, it became increasingly apparent through the development of our guidance that this theory item could relate to either existing published theory or programme theory	Chapters 2 & 5	
7. Report any use of components from an existing intervention in the current intervention development process.	Some interventions are developed with components that have been adopted from existing interventions. Clearly identifying components that have been adopted or adapted and acknowledging their original source helps the reader to understand and distinguish between the novel and adopted components of the new intervention.	N/A	
8. Report any guiding principles, people or factors that were prioritised when making decisions during the intervention development process.	Reporting any guiding principles that governed the development of the application helps the reader to understand the authors' reasoning behind the decisions that were made. These could include the examples of particular populations who views are being considered when designing the intervention, the modality that is viewed as being most appropriate, design features considered important for the target population, or the potential for the intervention to be scaled up.	Chapters 1, 2 & 5	

Item description	Explanation	Page in manuscript where item is located	Other*
9. Report how stakeholders contributed to the intervention development process.	Potential stakeholders can include patient and community representatives, local and national policy makers, health care providers and those paying for or commissioning health care. Each of these groups may influence the intervention development process in different ways. Specifying how differing groups of stakeholders contributed to the intervention development process helps the reader to understand how stakeholders were involved and the degree of influence they had on the overall process. Further detail on how to integrate stakeholder contributions within intervention reporting are available (19).	Chapters 1, 2, 3, 4 & 5	
10. Report how the intervention changed in content and format from the start of the intervention development process.	Intervention development is frequently an iterative process. The conclusion of the initial phase of intervention development does not necessarily mean that all uncertainties have been addressed. It is helpful to list remaining uncertainties such as the intervention intensity, mode of delivery, materials, procedures, or type of location that the intervention is most suitable for. This can guide other researchers to potential future areas of research and practitioners about uncertainties relevant to their healthcare context.	Chapter 5	
11. Report any changes to interventions required or likely to be required for subgroups.	Specifying any changes that the intervention development team perceive are required for the intervention to be delivered or tailored to specific sub groups enables readers to understand the applicability of the intervention to their target population or context. These changes could include changes to personnel delivering the intervention, to the content of the intervention, or to the mode of delivery of the intervention.	Chapter 5	
12. Report important uncertainties at the end of the intervention development process.	Intervention development is frequently an iterative process. The conclusion of the initial phase of intervention development does not necessarily mean that all uncertainties have been addressed. It is helpful to list remaining uncertainties such as the intervention intensity, mode of delivery, materials, procedures, or type of location that the intervention is most suitable for. This can guide other researchers to potential future areas of research and practitioners about uncertainties relevant to their healthcare context.	Chapter 5 & 6	
13. Follow TIDieR guidance when describing the developed intervention.	Interventions have been poorly reported for a number of years. In response to this, internationally recognized guidance has been published to support the high quality reporting of health care? interventions[5]and public health interventions[14]. This guidance should therefore be followed when describing a developed intervention.	Chapter 5	
14. Report the intervention development process in an open access format.	Unless reports of intervention development are available people considering using an intervention cannot understand the process that was undertaken and make a judgement about its appropriateness to their context. It also limits cumulative learning about intervention development methodology and observed consequences at later evaluation, translation and implementation stages. Reporting intervention development in an open access (Gold or Green) publishing format increases the accessibility and visibility of intervention development research and makes it more likely to be read and used. Potential platforms for open access publication of intervention development include open access journal publications, freely accessible funder reports or a study web-page that details the intervention development process.	In press	

*e.g. if item is reported elsewhere, then the location of this information can be stated here.

Appendix O – App-testing participant information sheet: child

UNIVERSITY OF LEEDS

NON-STOP: NON-Surgical Treatment Of Perthes' Disease

Chief Investigator(s):

Professor Anthony Redmond and Mr Adam Galloway

INFORMATION SHEET FOR CHILDREN

BACKGROUND

You have been invited to take part in the NON-STOP study because you have Perthes' Disease. The NON-STOP study is looking at an app to help us look after boys and girls like you with Perthes' Disease. It has some physiotherapy exercises for you to do and another page where we can tell you more about Perthes' Disease.

You and your guardian/carer said that we could talk to you about the app a bit more.

We want you to use the app for six weeks so we can see whether it is good or not.

There are a few more things we need to tell you.

DO I HAVE TO SAY YES?

No. If you say no, that's okay and it won't change how people in the hospital look after you.

WHAT HAPPENS IF I SAY YES?

We will tell you how to download and use the app. You will be able to ask us questions about the app too.

This might be at home or at school, and you'll use the app on a tablet or phone.

You and your parent/carer will answer some questions before you use start using the app, and again at the end of the six weeks.

WHO WILL KNOW WHO I AM IN THE STUDY?

Only people doing the research will know who you are in the study.

WHAT HAPPENS IF I WANT TO STOP?

You can stop talking whenever you want to, and we will stop.

WHAT WILL HAPPEN TO THE RESULTS OF THE STUDY?

We will send you and your family the results of the study and tell you what we found out after we have finished.

Thank you

Appendix P – App-testing participant information sheet: family

UNIVERSITY OF LEEDS

NON-STOP: NON-Surgical Treatment Of Perthes' Disease

Chief Investigator(s):

Professor Anthony Redmond and Mr Adam Galloway

INFORMATION SHEET FOR PARENT/GUARDIAN

BACKGROUND

You have been invited to take part in the NON-STOP study because your child has Perthes' Disease. The NON-STOP study is aiming to test a new app to help children with Perthes' Disease and their families, such as yourself, manage their condition. Previously you agreed we could contact you to discuss taking part in a research study. The aim of the study is to monitor how the app is used over a six-week period.

This document will explain in more detail what the study will include. **Please take time to read this and feel free to discuss with others if you wish.**

WHAT HAPPENS IF I SAY YES?

If you and your child decide to take part in the study, we will ask you to reply to the email that contained this sheet, with an 'agreement statement' as described, this will act as consent to take part in the study. It will say that you have read this information sheet, had time to decide whether to be involved and agree to take part. We will then arrange to set you up with the NON-STOP app and give you instructions on how to use it over the six weeks.

At the beginning of the study, you'll be asked to provide some information relating to your child's condition, then you'll be instructed to download the app. During the six-week period you'll use the app at home with your child. The app includes information about Perthes' Disease as well as instructional videos for children on how to complete their physiotherapy exercises.

After the six weeks have passed, you'll be asked to provide some more information about your child's condition again and instructed that this is the end of the study period. At this point you'll still have access to the app, but we will stop collecting data.

DO I HAVE TO TAKE PART?

No. It is completely up to you whether you would like to take part, and you do not need to decide straight away. You are also free to change your mind and withdraw from the study at any point. It is also absolutely fine if your child doesn't want to take part. Whether you/your child decide to take part or not will not affect the clinical care that your child receives.

ARE THERE ANY BENEFITS TO TAKING PART?

There are no specific benefits to taking part in the study. However you will have the opportunity to use a new treatment approach in the care of Perthes' Disease. Also, you'd be contributing to a study that could change the way we care for children with Perthes' Disease.

ARE THERE ANY RISKS IN TAKING PART?

Similar to the benefits, there are no specific risks in taking part. There is a time-burden involved with taking part, but we hope that this is minimal and that you can use the app when you have time.

HOW WILL WE USE INFORMATION FROM YOU?

We will need to use information from you for this research project. This information will include contact details [name, email address and phone number]. People will use this information to do the research or to check your records to make sure that the research is being done properly.

We will keep all information about you safe and secure. Your details will be kept in password-protected files. Only the research study team will have access to this. People who do not need to know who you are will not be able to see your name or contact details. Your data will have a code number instead. After the study you will be sent a summary of the research study and then your details will be destroyed.

Once we have finished the study, we will keep some of the data so we can check the results. We will write our reports in a way that no-one can work out that you took part in the study.

If you have any concerns about data privacy during the study you can email dpo@leeds.ac.uk

Alternatively, visit https://dataprotection.leeds.ac.uk/wp-content/uploads/sites/48/2020/08/My_data_and_research.pdf , https://dataprotection.leeds.ac.uk/wp-content/uploads/sites/48/2019/02/Research-Privacy-Notice.pdf for more information on data privacy.

WHAT WILL HAPPEN TO THE RESULTS OF THE STUDY?

You will receive a summary of the study once we have analysed the results. There will be reports published in medical journals and at conferences, that will be available to you if you wish. We will also provide a summary of the research findings on social media pages and relevant Perthes' Disease charity pages. This piece of research is being done as part of a PhD; the results will contribute to the thesis written as part of this.

WHO HAS REVIEWED THIS STUDY?

All research in the NHS is looked at by an independent group of people, called a Research Ethics Committee, to protect your interests. This study has been reviewed and given favourable opinion by the Edgbaston Research Ethics Committee.

WHAT IF THERE IS A PROBLEM?

This study is sponsored by the University of Leeds. If you wish to discuss any aspect of the research study then you can contact the Chief Investigator, Professor Anthony Redmond at a.redmond@leeds.ac.uk. For complaints, contact your local PALS on 0113 2066261.

You can stop being part of the study at any time, without giving a reason, but we will keep information about you that we already have.

We need to manage your records in specific ways for the research to be reliable. This means that we won't be able to let you see or change the data we hold about you.

FURTHER INFORMATION AND CONTACT DETAILS

You can find out more about how we use your information:

- at www.hra.nhs.uk/information-about-patients/
- by asking one of the research team by sending an email to a.galloway@leeds.ac.uk

Thank you for taking the time to read this information and consider taking part in our study.

Appendix Q – Focus group participant information sheet: child

UNIVERSITY OF LEEDS

NON-STOP: NON-Surgical Treatment Of Perthes' Disease

Chief Investigator(s):

Professor Anthony Redmond and Mr Adam Galloway

INFORMATION SHEET FOR CHILDREN

BACKGROUND

You have been invited to take part in the NON-STOP study because you have Perthes' Disease. The NON-STOP study is looking at an app to help us look after boys and girls like you with Perthes' Disease which you have been using.

You and your guardian/carer said that we could talk to you about the app a bit more.

We want to know what you have liked about using the app and what you have not liked as much.

There are a few more things we need to tell you.

DO I HAVE TO SAY YES?

No. If you say no, that's okay and it won't change how people in the hospital look after you.

WHAT HAPPENS IF I SAY YES?

We will ask you to come and talk to us in person about the app. There will be 3-4 other children with Perthes' Disease there who have used the app too.

Your guardian/carer will be with you the whole time.

You can ask any questions about the study.

We will ask you some questions about the app, to try and find out what you and the other children thought of the app.

We'll record the group so we can remember them later.

It will take about 20 minutes for us to talk, but you can stop whenever you want.

WHO WILL KNOW WHO I AM IN THE STUDY?

Only people doing the research will know who you are in the study.

WHAT HAPPENS IF I WANT TO STOP?

You can stop talking whenever you want to, and we will stop.

WHAT WILL HAPPEN TO THE RESULTS OF THE STUDY?

We will send you and your family the results of the study and tell you what we found out after we have finished.

Thank you

Appendix R – Focus group participant information sheet: family

UNIVERSITY OF LEEDS

NON-STOP: NON-Surgical Treatment Of Perthes' Disease

Chief Investigator(s):

Professor Anthony Redmond and Mr Adam Galloway

INFORMATION SHEET FOR PARENT/GUARDIAN

BACKGROUND

You have been invited to take part in the NON-STOP study because your child has Perthes' Disease and you recently tested the NON-STOP app. The NON-STOP study is aiming to test a new app to help children with Perthes' Disease and their families, such as yourself, manage their condition. Previously you took part in the study to test this app and agreed we could contact you to discuss you and your child taking part in a further study as part of the app-testing. The aim of the study is to ask a small group of people their thoughts on the app and experiences of using it.

This document will explain in more detail what the study will include. **Please take time to read this and feel free to discuss with others if you wish.**

WHAT HAPPENS IF I SAY YES?

If you decide to take part, we will ask you to reply to the email that contained this sheet, with an 'agreement statement' as described, this will act as consent for you and your child to take part in the study. It will say that you have read this information sheet, had time to decide whether to be involved and agree to take part. You and your child will then be invited to a face-to-face group interview called a focus group at the hospital you go to for your appointments about Perthes' Disease. There will be a group of 4-5 children in one group discussing their experiences of using the NON-STOP app. After this group has completed, a group of 4-5 adults will discuss their experiences of using the NON-STOP app.

The focus group will take between 30-90 minutes in total and will be recorded using a small recording device. An external transcription service will type up the focus groups so that the researcher can look at your answers again at a later time. There is a confidentiality agreement in place between the external company and University of Leeds to make sure your information is safe. None of your personal details will be included in the transcription. Once we've typed them up, we'll destroy the audio recordings, but the typed-up answers will be kept in password-protected files on University of Leeds computers for 10 years, in line with GDPR guidance, after this they'll be destroyed.

DO I HAVE TO TAKE PART?

No. It is completely up to you whether you would like to take part, and you do not need to decide straight away. You are also free to change your mind and withdraw from the study at any point. Whether you/your child decide to take part or not will not affect the clinical care that your child receives.

UNIVERSITY OF LEEDS

ARE THERE ANY BENEFITS TO TAKING PART?

There are no specific benefits to taking part in the interview. However sometimes people feel there is a benefit to sharing their experiences and feel that being involved in research studies like this can help contribute to a better understanding of the condition. This might be particularly important in a condition like Perthes' Disease where we don't know an awful lot about the experiences of those most involved.

ARE THERE ANY RISKS IN TAKING PART?

Similar to the benefits, there are no specific risks in taking part. It is possible that having to recall previous experiences could bring back memories of something potentially upsetting. The research team will be able provide support however, and offer the chance to take a break or, if needed, stop the focus group at any point.

In the rare occasion there are any safeguarding issues, confidential information may need to be shared with your existing medical team.

We appreciate that it can be time-consuming to take part in research, and because of this, we have funding to reimburse you and your child for your time and have a childcare allowance if this makes it more realistic for you to take part. In line with NIHR guidance this will be a maximum of £50.

HOW WILL WE USE INFORMATION FROM YOU?

We will need to use information from you for this research project. This information will include contact details [name, email address and phone number]. People will use this information to do the research or to check your records to make sure that the research is being done properly.

We will keep all information about you safe and secure. Your details will be kept in password-protected files. Only the research study team will have access to this. After the study you will be sent a summary of the research study and then your details will be destroyed.

Once we have finished the study, we will keep some of the data so we can check the results. We will write our reports in a way that no-one can work out that you took part in the study.

If you have any concerns about data privacy during the study you can email dpo@leeds.ac.uk

Alternatively, you can visit https://dataprotection.leeds.ac.uk/wp-content/uploads/sites/48/2020/08/My_data_and_research.pdf , https://dataprotection.leeds.ac.uk/wp-content/uploads/sites/48/2019/02/Research-Privacy-Notice.pdf or HRA website www.hra.nhs.uk/information-about-patients/

for more information on data privacy.

UNIVERSITY OF LEEDS

WHAT WILL HAPPEN TO THE RESULTS OF THE STUDY?

As mentioned previously, you will receive a summary of the study once we have analysed the focus group. There will be reports published in medical journals and at conferences, that will be available to you if you wish, but these will not name any participants. We will also provide a summary of the research findings on social media pages and relevant Perthes' Disease charity pages.

This piece of research is being done as part of a PhD; the results will contribute to the thesis written as part of this.

WHO HAS REVIEWED THIS STUDY?

All research in the NHS is looked at by an independent group of people, called a Research Ethics Committee, to protect your interests. This study has been reviewed and given favourable opinion by the Edgbaston Research Ethics Committee.

WHAT IF THERE IS A PROBLEM?

This study is sponsored by the University of Leeds. If you wish to discuss any aspect of the research study then you can contact the Chief Investigator, Professor Anthony Redmond at a.redmond@leeds.ac.uk. For complaints, contact your local PALS on 0113 2066261.

You can stop being part of the study at any time, without giving a reason, but we will keep information about you that we already have.

We need to manage your records in specific ways for the research to be reliable. This means that we won't be able to let you see or change the data we hold about you.

FURTHER INFORMATION AND CONTACT DETAILS

You can find out more about how we use your information:

- at www.hra.nhs.uk/information-about-patients/
- by asking one of the research team by sending an email to a.galloway@leeds.ac.uk

Thank you for taking the time to read this information and consider taking part in our study.

Appendix S – CONSORT checklist for feasibility study

CONSORT 2010 checklist of information to include when reporting a pilot or feasibility trial*

Section/Topic	Item No	Checklist item	Reported on page No
Title and abstract			
	1a	Identification as a pilot or feasibility randomised trial in the title	179
	1b	Structured summary of pilot trial design, methods, results, and conclusions (for specific guidance see CONSORT abstract extension for pilot trials)	N/A
Introduction			
Background and objectives	2a	Scientific background and explanation of rationale for future definitive trial, and reasons for randomised pilot trial	192
	2b	Specific objectives or research questions for pilot trial	179/180
Methods			
Trial design	3a	Description of pilot trial design (such as parallel, factorial) including allocation ratio	N/A
	3b	Important changes to methods after pilot trial commencement (such as eligibility criteria), with reasons	N/A
Participants	4a	Eligibility criteria for participants	183
	4b	Settings and locations where the data were collected	184-186
	4c	How participants were identified and consented	184-190
Interventions	5	The interventions for each group with sufficient details to allow replication, including how and when they were actually administered	197
Outcomes	6a	Completely defined prespecified assessments or measurements to address each pilot trial objective specified in 2b, including how and when they were assessed	192-200
	6b	Any changes to pilot trial assessments or measurements after the pilot trial commenced, with reasons	N/A
	6c	If applicable, prespecified criteria used to judge whether, or how, to proceed with future definitive trial	N/A
Sample size	7a	Rationale for numbers in the pilot trial	183/184
	7b	When applicable, explanation of any interim analyses and stopping guidelines	N/A
Randomisation:			
Sequence generation	8a	Method used to generate the random allocation sequence	N/A
	8b	Type of randomisation(s); details of any restriction (such as blocking and block size)	N/A
Allocation concealment mechanism	9	Mechanism used to implement the random allocation sequence (such as sequentially numbered containers), describing any steps taken to conceal the sequence until interventions were assigned	N/A

Implementation	10	Who generated the random allocation sequence, who enrolled participants, and who assigned participants to interventions	N/A
Blinding	11a	If done, who was blinded after assignment to interventions (for example, participants, care providers, those assessing outcomes) and how	N/A
	11b	If relevant, description of the similarity of interventions	N/A
Statistical methods	12	Methods used to address each pilot trial objective whether qualitative or quantitative	200-209
Results			
Participant flow (a diagram is strongly recommended)	13a	For each group, the numbers of participants who were approached and/or assessed for eligibility, randomly assigned, received intended treatment, and were assessed for each objective	211
	13b	For each group, losses and exclusions after randomisation, together with reasons	211
Recruitment	14a	Dates defining the periods of recruitment and follow-up	211
	14b	Why the pilot trial ended or was stopped	N/A
Baseline data	15	A table showing baseline demographic and clinical characteristics for each group	211/212
Numbers analysed	16	For each objective, number of participants (denominator) included in each analysis. If relevant, these numbers should be by randomised group	212
Outcomes and estimation	17	For each objective, results including expressions of uncertainty (such as 95% confidence interval) for any estimates. If relevant, these results should be by randomised group	N/A
Ancillary analyses	18	Results of any other analyses performed that could be used to inform the future definitive trial	N/A
Harms	19	All important harms or unintended effects in each group (for specific guidance see CONSORT for harms)	N/A
	19a	If relevant, other important unintended consequences	N/A
Discussion			
Limitations	20	Pilot trial limitations, addressing sources of potential bias and remaining uncertainty about feasibility	235
Generalisability	21	Generalisability (applicability) of pilot trial methods and findings to future definitive trial and other studies	235
Interpretation	22	Interpretation consistent with pilot trial objectives and findings, balancing potential benefits and harms, and considering other relevant evidence	240
	22a	Implications for progression from pilot to future definitive trial, including any proposed amendments	240/241
Other information			
Registration	23	Registration number for pilot trial and name of trial registry	269
Protocol	24	Where the pilot trial protocol can be accessed, if available	N/A
Funding	25	Sources of funding and other support (such as supply of drugs), role of funders	209
	26	Ethical approval or approval by research review committee, confirmed with reference number	209

Appendix T – Children's Physical Activity Questionnaire

CHILDREN'S PHYSICAL ACTIVITY QUESTIONNAIRE (C-PAQ)

Parent Questionnaire

Your child's name: .....

Your child's date of birth (dd/mm/yy): ..... / ..... / .....

Are you the child's: mother / father / guardian / other

Please note: - this questionnaire will take approximately 10 minutes to complete
- please answer the questions in relation to the child named above
- please **complete every line** in the questionnaire

For further information, please contact:

Which of the following PHYSICAL activities did your child do in the PAST 7 DAYS?

Please complete this questionnaire for the following days: …… to ……

Did your CHILD do the following activities in the **past 7 days?**			**MONDAY – FRIDAY**		**SATURDAY – SUNDAY**	
			How many times **Mon–Fri**?	**Total** hours/minutes **Mon-Fri**?	How many times **Sat-Sun**?	**Total** hours/minutes **Sat-Sun**?
EXAMPLE: **Bike riding**	**No**	**Yes**	**2**	**40 mins**	**1**	**15 mins**
SPORTS ACTIVITIES Aerobics	No	Yes				
Baseball/softball	No	Yes				
Basketball/volleyball	No	Yes				
Cricket	No	Yes				
Dancing	No	Yes				
Football	No	Yes				
Gymnastics	No	Yes				
Hockey (field or ice)	No	Yes				
Martial arts	No	Yes				
Netball	No	Yes				
Rugby	No	Yes				

Did your CHILD do the following activities in the **past 7 days?**			**MONDAY – FRIDAY**		**SATURDAY – SUNDAY**	
			How many times **Mon–Fri**?	**Total** hours/minutes **Mon-Fri**?	How many times **Sat-Sun**?	**Total** hours/minutes **Sat-Sun**?
Running or jogging	No	Yes				
Swimming lessons	No	Yes				
Swimming for fun	No	Yes				
Tennis/badminton/squash/ other racquet sport	No	Yes				
LEISURE TIME ACTIVITIES Bike riding (not school travel)	No	Yes				
Bounce on the trampoline	No	Yes				
Bowling	No	Yes				
Household chores	No	Yes				
Play in a play house	No	Yes				
Play on playground equipment	No	Yes				
Play with pets	No	Yes				
Rollerblading/roller-skating	No	Yes				
Scooter	No	Yes				

Did your CHILD do the following activities in the **past 7 days?**			**MONDAY – FRIDAY**		**SATURDAY – SUNDAY**	
			How many times **Mon–Fri**?	**Total** hours/minutes **Mon-Fri**?	How many times **Sat-Sun**?	**Total** hours/minutes **Sat-Sun**?
Skateboarding	No	Yes				
Skiing, snowboarding, sledging	No	Yes				
Skipping rope	No	Yes				
Tag	No	Yes				
Walk the dog	No	Yes				
Walk for exercise/hiking	No	Yes				
ACTIVITIES AT SCHOOL Physical education class	No	Yes				
Travel by walking to school (to and from school = 2 times)	No	Yes				
Travel by cycling to school (to and from school = 2 times)	No	Yes				
OTHER please state:	No	Yes				

Did your CHILD do the following activities in the **past 7 days?**		**MONDAY-FRIDAY** **Total** hours/minutes	**SATURDAY-SUNDAY** **Total** hours/minutes
EXAMPLE: **Watching TV/videos**	**No** **Yes**	**15hrs**	**6hrs 30mins**
Art & craft (eg. pottery, sewing, drawing, painting)	No Yes		
Doing homework	No Yes		
Imaginary play	No Yes		
Listen to music	No Yes		
Play indoors with toys	No Yes		
Playing board games / cards	No Yes		
Playing computer games (e.g. playstation / gameboy)	No Yes		
Playing musical instrument	No Yes		
Reading	No Yes		
Sitting talking	No Yes		
Talk on the phone	No Yes		
Travel by car / bus to school (to and from school)	No Yes		

Did your CHILD do the following activities in the **past 7 days?**		**MONDAY-FRIDAY** **Total** hours/minutes	**SATURDAY-SUNDAY** **Total** hours/minutes
Using computer / internet	No Yes		
Watching TV/videos	No Yes		
Other (please state):	No Yes		

Appendix U – Health ITUES for the NON-STOP app

Health-IT Usability Evaluation Scale (Health ITUES) for the NON-STOP app

Response options for all items are: Strongly Agree, Somewhat Agree, Neither Agree nor Disagree, Somewhat Disagree, and Strongly Disagree.

Impact

1. I think the NON-STOP app would benefit persons living with Perthes' Disease.
2. I think the NON-STOP app would improve the quality of life of persons living with Perthes' Disease.
3. The NON-STOP app is an important part of meeting my information needs related to my health.

Perceived usefulness

4. Using The NON-STOP app will make it easier for me to monitor and learn about my health.
5. Using The NON-STOP app will enable me to monitor and learn about my health.
6. Using the NON-STOP app makes it more likely that I will track my health.
7. Using the NON-STOP app will be useful for receiving reminders about my health.
8. I think the NON-STOP app presents a more equitable process for managing my health.
9. I am satisfied with the NON-STOP app for helping me monitor and learn more about my health.
10. I will monitor my health in a timely manner because of the NON-STOP app.
11. Using the NON-STOP app will increase my ability to track my health.
12. I will be able to track my health whenever I use the NON-STOP app.

Perceived ease of use

13. I am comfortable with my ability to use the NON-STOP app.
14. Learning to operate the NON-STOP app is easy for me.
15. It will be easy for me to become skillful at using the NON-STOP app.
16. I find the NON-STOP app easy to use.
17. I can always remember how to log on to and use The NON-STOP app.

User control

18. The NON-STOP app gives error messages that clearly tell me how to fix problems.
19. Whenever I make a mistake using the NON-STOP app, I recover easily and quickly.
20. The information (such as on-line help, on-screen messages and other documentation) provided with the NON-STOP app is clear.

Appendix V – Topic guide for focus group

NON-STOP: NON-Surgical Treatment Of Perthes' Disease

Chief Investigator(s):

Professor Anthony Redmond and Mr Adam Galloway

TOPIC GUIDE FOR CHILD/FAMILY

AIM OF THIS GUIDE

This guide is for the facilitator to use in order to guide the focus group with the participants.

It includes some questions and prompts that can be used during the focus group as well as reminders for the facilitator as to the format of the interview.

IMPORTANT STEPS PRIOR TO STARTING INTERVIEW

Ensure participants have had time to read and understand the participant information pack and any further check for any further questions they may have. Ensure participants understand that they do not have to take part in the focus group and that they can stop at any time.

At the beginning of the focus group, make it clear that the group will be recorded but that everything said will remain confidential and any information used will be anonymised. Their clinical care team will not be informed about any of the answers that they give in the discussions. Be sure to inform the participant that recording has started and stopped.

GROUND RULES

Respect each other's space & try not to talk over each other

This is a confidential and safe space, please don't repeat what is said

QUESTIONS/PROMPTS TO BE USED DURING INTERVIEW

Main questions/aims:

1. What did you think of the NON-STOP app? (aim to get responses around ease of use, when they used it, etc.)
2. How was using it compared to your existing care? (think this is where the prompt for reminders would be, and phrased "how did you find the reminders built in to the app?")
3. What would you change about the app? (to get some clear ideas for the future)

Prompts (Child):

- What did you think of using the app?
- What happened when you tried to load it up?
- What parts of the app did you like? And why?
- What parts of the app did you not like so much? And why?
- How did using the app fit in with your life?
- How did using the app compare to doing your normal physiotherapy?

Prompts (Parent/legal guardian):

- What are your experiences of you/your child using the app?
- How did using the app compare to your normal routine?
- What are your thoughts about using an app like this for long-term self-management?
- Ask child/family if they have any questions or if there is something else that they would like to add.

FINAL ACTIONS

Reiterate the plan following focus i.e. additional groups with other participants, data analysis and the dissemination plan.

"How have you found the focus group" – as a transition 'out' of the study.

Appendix W – COREQ checklist for nested qualitative study

COREQ (COnsolidated criteria for REporting Qualitative research) Checklist

A checklist of items that should be included in reports of qualitative research. You must report the page number in your manuscript where you consider each of the items listed in this checklist. If you have not included this information, either revise your manuscript accordingly before submitting or note N/A.

Topic	Item No.	Guide Questions/Description	Reported on Page No.
Domain 1: Research team and reflexivity			
Personal characteristics			
Interviewer/facilitator	1	Which author/s conducted the interview or focus group?	198
Credentials	2	What were the researcher's credentials? E.g. PhD, MD	198
Occupation	3	What was their occupation at the time of the study?	198
Gender	4	Was the researcher male or female?	n/a
Experience and training	5	What experience or training did the researcher have?	198
Relationship with participants			
Relationship established	6	Was a relationship established prior to study commencement?	N/A
Participant knowledge of the interviewer	7	What did the participants know about the researcher? e.g. personal goals, reasons for doing the research	N/A
Interviewer characteristics	8	What characteristics were reported about the inter viewer/facilitator? e.g. Bias, assumptions, reasons and interests in the research topic	199
Domain 2: Study design			
Theoretical framework			
Methodological orientation and Theory	9	What methodological orientation was stated to underpin the study? e.g. grounded theory, discourse analysis, ethnography, phenomenology, content analysis	203
Participant selection			
Sampling	10	How were participants selected? e.g. purposive, convenience, consecutive, snowball	182
Method of approach	11	How were participants approached? e.g. face-to-face, telephone, mail, email	185
Sample size	12	How many participants were in the study?	217
Non-participation	13	How many people refused to participate or dropped out? Reasons?	217
Setting			
Setting of data collection	14	Where was the data collected? e.g. home, clinic, workplace	198
Presence of non-participants	15	Was anyone else present besides the participants and researchers?	198
Description of sample	16	What are the important characteristics of the sample? e.g. demographic data, date	198
Data collection			
Interview guide	17	Were questions, prompts, guides provided by the authors? Was it pilot tested?	216
Repeat interviews	18	Were repeat inter views carried out? If yes, how many?	N/A
Audio/visual recording	19	Did the research use audio or visual recording to collect the data?	198
Field notes	20	Were field notes made during and/or after the inter view or focus group?	198
Duration	21	What was the duration of the inter views or focus group?	199
Data saturation	22	Was data saturation discussed?	N/A
Transcripts returned	23	Were transcripts returned to participants for comment and/or	N/A

Topic	Item No.	Guide Questions/Description	Reported on Page No.
		correction?	
Domain 3: analysis and findings			
Data analysis			
Number of data coders	24	How many data coders coded the data?	205
Description of the coding tree	25	Did authors provide a description of the coding tree?	207
Derivation of themes	26	Were themes identified in advance or derived from the data?	205
Software	27	What software, if applicable, was used to manage the data?	209
Participant checking	28	Did participants provide feedback on the findings?	N/A
Reporting			
Quotations presented	29	Were participant quotations presented to illustrate the themes/findings? Was each quotation identified? e.g. participant number	217-232
Data and findings consistent	30	Was there consistency between the data presented and the findings?	N/A
Clarity of major themes	31	Were major themes clearly presented in the findings?	207
Clarity of minor themes	32	Is there a description of diverse cases or discussion of minor themes?	232

Developed from: Tong A, Sainsbury P, Craig J. Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *International Journal for Quality in Health Care*. 2007. Volume 19, Number 6: pp. 349 – 357

Once you have completed this checklist, please save a copy and upload it as part of your submission. DO NOT include this checklist as part of the main manuscript document. It must be uploaded as a separate file.

Appendix X – HRA/REC favourable opinion letter for feasibility study

Professor Anthony Redmond
Leeds Institute of Theumatic and Musculoskeletal Medicine
School of Medicine, University of Leeds
Level 2, Chapel Allerton Hospital, Leeds
LS7 4SA

Email: approvals@hra.nhs.uk
HCRW.approvals@wales.nhs.uk

30 November 2023

Dear Professor Redmond

HRA and Health and Care Research Wales (HCRW) Approval Letter

Study title:	**Evaluating the acceptability and usability of a digital self-management intervention to support the non-surgical treatment of Perthes' Disease**
IRAS project ID:	**330507**
Protocol number:	**1**
REC reference:	**23/WM/0251**
Sponsor	**University of Leeds**

I am pleased to confirm that **HRA and Health and Care Research Wales (HCRW) Approval** has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications received. You should not expect to receive anything further relating to this application.

Please now work with participating NHS organisations to confirm capacity and capability, in line with the instructions provided in the "Information to support study set up" section towards the end of this letter.

How should I work with participating NHS/HSC organisations in Northern Ireland and Scotland?
HRA and HCRW Approval does not apply to NHS/HSC organisations within Northern Ireland and Scotland.

If you indicated in your IRAS form that you do have participating organisations in either of these devolved administrations, the final document set and the study wide governance report (including this letter) have been sent to the coordinating centre of each participating nation. The relevant national coordinating function/s will contact you as appropriate.

Please see IRAS Help for information on working with NHS/HSC organisations in Northern Ireland and Scotland.

How should I work with participating non-NHS organisations?
HRA and HCRW Approval does not apply to non-NHS organisations. You should work with your non-NHS organisations to obtain local agreement in accordance with their procedures.

What are my notification responsibilities during the study?

The standard conditions document "*After Ethical Review – guidance for sponsors and investigators*", issued with your REC favourable opinion, gives detailed guidance on reporting expectations for studies, including:

- Registration of research
- Notifying amendments
- Notifying the end of the study

The HRA website also provides guidance on these topics, and is updated in the light of changes in reporting expectations or procedures.

Who should I contact for further information?
Please do not hesitate to contact me for assistance with this application. My contact details are below.

Your IRAS project ID is **330507**. Please quote this on all correspondence.

Yours sincerely,
Holly Lloyd

Approvals Specialist

Email: approvals@hra.nhs.uk

Copy to: *Mrs Jean Uniacke*

List of Documents

The final document set assessed and approved by HRA and HCRW Approval is listed below.

Document	*Version*	*Date*
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [Indemnity]		25 October 2023
Interview schedules or topic guides for participants [Topic guide]	1	25 October 2023
IRAS Application Form [IRAS_Form_30102023]		30 October 2023
IRAS Application Form XML file [IRAS_Form_30102023]		30 October 2023
IRAS Checklist XML [Checklist_30102023]		30 October 2023
Organisation Information Document [OID]	1	26 October 2023
Other [CPAQ]	1	25 October 2023
Other [IRAS changes]	1	22 November 2023
Other [IRAS changes]	2	23 November 2023
Other [Revised invite]	2	23 November 2023
Other [Revised invite]	2	23 November 2023
Other [Revised PIS App-testing child]	3	23 November 2023
Other [Revised PIS focus group child]	3	23 November 2023
Participant information sheet (PIS) [PIS Qual Family]	4	30 November 2023
Participant information sheet (PIS) [PIS App Family]	4	30 November 2023
Research protocol or project proposal [Protocol]	1	25 October 2023
Schedule of Events or SoECAT [SoECAT]	1	25 October 2023
Summary CV for Chief Investigator (CI) [ACR CV]	1	30 October 2023
Summary CV for student [AG CV]		25 October 2023
Summary CV for supervisor (student research) [ACR CV]		25 October 2023
Summary of any applicable exclusions to sponsor insurance (non-NHS sponsors only) [Indemnity]	1	25 October 2023
Validated questionnaire [PROM]		

Appendix Y – Participant quote table from focus groups

Quote table for qualitative study

Code	Theme	Participant quoting
1	**Ease of use**	
1.1	Problems with use	
1	*if I was working or he was, you couldn't use it on different devices. Dad had it on his phone but then when he went on, he had to restart. If child already had, say, three stars that week, it didn't connect.*	Mother of 4-year-old male
	there was also a couple of times where I had gone on and done it with him but there must have been, I don't know, something to do with the technology of it, that it didn't load until the next day so then we couldn't go on again	Mother of 4-year-old male
2	*Sometimes they (the app reminders) came through, sometimes they didn't*	Mother of 5-year-old male
	There were a couple of issues where we'd done the exercises and then it just refreshed. And then when you've already done the exercises, as if it's gone back a day.	Mother of 5-year-old male
	It was playing up on my phone for a week or so, but it seems to be absolutely fine, now. I think it's just probably because I had a new phone.	Father of 7-year-old male
1	*It doesn't match up (sessions on different phones with same log in). Mum has more data on her phone than mine*	Father of 7-year-old male
1.2	Elements increasing use	
	with the exercises on the app, we find that we can just do them all together. He's even got my mum and dad doing them.	Mother of 5-year-old male
1	*the app was showing you how to do it. Again, I felt like it was more of a game for him, it was more entertaining, it was like a race, how many can you do before the time runs out*	Mother of 5-year-old male
	Even just getting the sticker, he would be happy but he is at that age where he's getting stars. He's like, I got another star, I got this, I got that. He was going back telling my parents, he was proud of that accomplishment	Mother of 5-year-old male
1	*I liked copying the exercises*	7-year-old male
	Father: It's like, it seems to be anything that's, like, gamified works *Child: Yeah* *Dad: helps, doesn't it?*	7-year-old male/father

1.3	Demonstration of independence	
	I think it's really allowed (child) to be aware of his Perthes' because we hadn't actually really told him. And then when the app came around, (child) is now more aware, he's only four, and it's allowed Albert to take control of it. He's able to say if he thinks it's hurting and for us to know more	Mother of 4-year-old male
	It's really child friendly for that and really accessible for even a four-year-old to click it, press it. He used to press which rating, choose his activities	Mother of 4-year-old male
2	*I feel like it really allowed me and Carl to really get an understanding of Perthes'*	Mother of 4-year-old male
	there are children of (child's) age who have Perthes', and it's good for them to be able to understand that they have something but it's manageable, they can do something to help it. And (child) was really excited coming this morning, as we came, he was like, we're not going to be late, are we, because I need to talk about my hip. And I thought, how good is that, at the age of four to say that	Mother of 4-year-old male
	with the ones on the app, we find that we can just do them altogether. He's even got my mum and dad doing them	Mother of 5-year-old male
	I do think it (the app) supports him a lot better. There are times where he's done the exercises himself	Mother of 5-year-old male
	The app has given him that independence	Mother of 5-year-old male
	(Child) would just take my phone, and she would go and sit and do it, she would get her sisters to join in, and stuff, it was great.	Mother of 7-year-old female
	Father of male: And you found it really straightforward, didn't you? He would go on, pain today, and then he has a little scroll to see, and off he goes. *Male, 7: yeah* *Mother of female: Yeah, that's the same as you isn't it* *Female, 7: Yeah*	Interaction between father of 7-year-old male and mother of 7-year-old female, and both children.
2	**Rewards**	
2.1	Impact of rewards on use	
	Mother: when you got your stars, what did you like to do with Bobby the Bone? *Child: Put things on him to make him look different*	Mother of 4-year-old male and child
3	*I think when we knew there were the stars, it's a goal for everybody and I think you need that in life, don't you, with everything*	Mother of 4-year-old male
	You liked getting the stars, didn't you, to be able to give Bobby the Bone different accessories. You liked to do that	Mother of 5-year-old male

2	*Even just getting the star, he would be happy, he is at that age where he's getting stars. He's like, "I got another star, I got this, I got that". He was going back telling my parents, he was proud of that accomplishment*	Mother of 5-year-old male
1	*Father: Why do you enjoy the app more? Because of Bobby, wasn't it? I think it was, the reward was getting to…* *Child: I like to customise Bobby the bone*	7-year-old male/father
	they were working towards something, and when they went on it, they got rewarded for doing it.	Father of 7-year-old male
	Father: It's like, it seems to be anything that's, like, gamified works *Child: Yeah* *Dad: helps, doesn't it?*	7-year-old male/father
	It's like, if you can match him with something that he enjoys…it's like the games factor then, isn't it.	Father of 7-year-old male
	(when describing struggling with motivation) *Father of male: It's not been like that with the app. But, yeah, it's kind of, it's getting them to buy into it, isn't it? And the animation, and the rewards with that, it seems to be working well.* *Mother of female: Yeah, that's a winner for us, the rewards.*	Interaction between father of 7-year-old male and mother of 7-year-old female
3	**Experience of the app**	
3.1	Positive elements of the app	
	I liked the butterfly	4-year-old male
	I think that it's really good that the app allows you to stop if you need to	Mother of 4-year-old male
	I do feel that this app really allowed me and (father), as I said, as a family, to really understand more about Perthes' because he only got diagnosed with it not so long ago	Mother of 4-year-old male
	I think it is in a really child-friendly way, even for the parents, and it's bright, it's colourful	Mother of 4-year-old male
	It was good but a lot of fun using	5-year-old male
	Mother: You liked getting the stars, didn't you, to be able to give Bobby the Bone different accessories. You liked to do that *Child: yeah*	Mother of 5-year-old male/child
	Mother: Scissor jumps and ninja squats. *Child: Because they make me better.*	Mother of 5-year-old male/child
2	*I thought it was absolutely brilliant, the way it was worded. There was more understanding because of the stuff that's online that you're trying to read up on is not very clear. And the way it described it as an ice cream, the way it melts and then. I thought, no-one's ever told me that*	Mother of 5-year-old male

	I think the app is a tremendous idea. One of the things that we did was…we did a load of personal trainer style videos for him that he would follow	Father of 6-year-old male
	I think the nice thing about the app is that it basically does that (provide exercises) on a professional level	Father of 6-year-old male
	the nice thing about the app is the progression. From the physio, you get a black and white printed out paper that says, this is what the exercise looks like, give it a go. Whereas actually the app is much more interactive and gives you a lot more to do	Father of 6-year-old male
1	*Female, 7: I think it's (the app) just good for everyone.* *Male, 7: Yeah. Really fun*	Interaction between 7-year-old male and female
	I like to customise Bobby the bone	7-year-old male
	I really liked the learning section, because obviously, we get a lot of, "well what is Perthes?", and I would just be like, "look, have a read"	Mother of 7-year-old female
	Father of male: the animation, and the rewards with that, it seems to be working well. *Mother of female: Yeah, that's a winner for us, the rewards.*	Interaction between father of 7-year-old male and mother of 7-year-old female
3.2	Less positive elements of the app	
1	*Three times a week, fine. I think when it got to the last week, I think they had to do it nearly every day*	Mother of 4-year-old male
2	*Sometimes they came through, sometimes they didn't*	Mother of 5-year-old male
3.3	Progression throughout app-testing period	
	I think the videos really helped; we watched the videos first. I think obviously as the weeks went on and it increased, we didn't need to watch the videos as much	Mother of 4-year-old male
	I don't know if that's because, obviously again, all the exercises he's doing and the confidence he has, but that was something he wouldn't have done a few weeks ago	Mother of 4-year-old male
	now, because he's missed doing things like football, and being able to go on things, he hasn't been able to, and he's kind of, well into that now, so it's kind of, a bit of a transition thing for him. But it's still keeping him doing his stretches, and his good movement	Father of 6-year-old male
4	**Comparison to previous/existing care**	
4.1	*Relationship to current care*	
1	*He was actually seeing (local physiotherapist) every two or three weeks, and now it's every four weeks*	Mother of 5-year-old male

2	*We do that including our physio, so we do both. The app is slightly different from the exercises she gets from physio*	Mother of 7-year-old female
4.2	Differences to previous care	
	I think he finds using the app more fun	Mother of 5-year-old male
1	*I definitely think he got more benefit from the app than us looking at a piece of paper and trying to figure it out, where the app was showing you how to do it.*	Mother of 5-year-old male
2	*he lost his hydrotherapy. We didn't get on with the physiotherapist, they said there was conflicting views, and things, and she was trying to influence the school, and things, and it went a bit crazy. So, Thomas has had some physio at another centre, but if we had the app earlier, it would have been really, really, really useful.*	Father of 7-year-old male
4.3	Relevance to stage of Perthes' Disease	
1	*The frog jump, yeah. I think a couple, he got better, but I think some of them, because of his age, some of them he probably didn't do as well as obviously somebody that's a bit older.*	Mother of 4-year-old male
	John wasn't that bothered about the rewards but then he's six and coming towards the end of it	Father of 6-year-old male
2	*now, because he's missed doing things like football, and being able to go on things, he hasn't been able to, and he's kind of, well into that now, so it's kind of, a bit of a transition thing for him. But it's still keeping him doing his stretches, and his good movement*	Father of 6-year-old male
5	**Future**	
5.1	Changes to functionality of the app	
2	*(when asked about potentially using videos in the Learning section)* *I would say, definitely for him, because he's reading but he wouldn't be able to read a full paragraph of what it's all explaining*	Mother of 5-year-old male
1	*I think if you've got two parents that are doing it together or you've got, say, an iPad and a phone, then I think it's probably good if that could work*	Mother of 4-year-old male
	It doesn't match up. She's got more data on her phone than mine. More stars, and things.	Father of 7-year-old male

5.2	Ideas for the app	
	(How would you dress Bobby up?) *In a football kit*	4-year-old male
	(Do you think the app would be good if it had more videos?) *Child: Yeah.* *(Who should be talking on the videos?)* *Child: Bobby The Bone*	4-year-old male
	I think for busy parents and busy family lives, I do think a reminder would be good	Mother of 4-year-old male
	Child: A pirate *Mother: Bobby the pirate*	4-year-old male and mother
	Change Bobby The Bone. Different things to give him. *(What would you like to see him dress up as?)* *A vampire.*	5-year-old male
1	*I would've liked to have something on where I could write in when he's had a bad day, so I could document it all in one area. And looking back on where it brings the faces on how he's feeling on that day, to go back and see what that is*	Mother of 5-year-old male
	A Halloween footballer. Easter Bunny and Santa.	5-year-old male
2	*different exercises as it progresses, he's a year in now*	Mother of 5-year-old male
	or as it's getting worse, are there different exercises that he would need to do as it's getting worse.	Father of 6-year-old male
	Would there be an option to do that longer or for more as the kid gets further on down the progression? For (child), what we did was repeated a lot of them	Father of 6-year-old male
	Father: Is there an overview of data that you can get? It might be worth if there's…at the end of the month if you can do a printout *Mother: It would be a good way of looking back again and seeing how when he was been able to complete it. Because there were times when he felt like he couldn't use it, like, Mum, I need to rest* *Father: that you can go through with the physio on the app, or you know, there's a PDF*	Interaction between father of 6-year-old male and mother of 5-year-old male
	(What would you like Bobby to dress up as?) *Female: a rock star* *Male: a rock star*	Interaction between 7-year-old female and male
	Father: Yeah, I think having videos in the learning section would be good, yeah *Mother: Yeah, it could be useful*	Father of 7-year-old male and mother of 7-year-old female
	You like your bridging, and stuff like that, don't you	Mother of 7-year-old female
	Father: in one physio session we had here, in Alder Hey, and they'd just got a little ball for him, and he was rolling, and making room into his, and he was happy, because it was a ball	Interaction between father of 7-year-old male and mother of 7-year-old female

	Mother: Yeah, Amelia's been doing that as well, she really enjoys it	
	I don't know how useful it would be to other parents, but we found that at the start of, at the onset of Perthes, is having little videos to show our Doctor. So, whether some video footage could be uploaded with the notes	Father of 7-year-old male
3	*you've got the QR code in the clinic, and I think that's got the guide on, hasn't it, of what the app is. That might be something you could share. Because that's where people are sat. You're sat, you're waiting, you've got your phone, obviously you're there for that reason on that day for your child with Perthes' and you're like, look, there's that. That, especially if parents haven't heard of it*	Mother of 4-year-old male
6	**AOB**	
6.1	Educating others	
	The school thought it was absolutely brilliant, the way it was explained. I was going to try and get them on the app as well, to try, because they will do his exercises with him as well	Mother of 5-year-old male
1	*I really liked that, because obviously, we get a lot of, well what is Perthes, and I would just be like, look, have a read, you know what I mean? Yeah, show the, like, school teachers, and like, obviously, family, and stuff like that, who would, yeah, like my sisters, and stuff like that, I'd be like, they'd be like, so what actually is it, and you'd be like, there you go, have a look at it, have a read.*	Mother of 7-year-old female
2	*it would have been useful for us to have that to show his school, and maybe they could have done, used the app themselves in school, when he was missing out on things in PE that he couldn't do, and whatnot*	Father of 7-year-old male
	I think, again, with the app, I think schools should, if there's children with Perthes in the school, I think they should have the access to the app as well.	Mother of 7-year-old female
	or when (child) is in doing PE, they've got, like, a big screen as well, where they do, like, yoga, and stuff like that. Well, they like to, obviously, get the other kids to join in with (child's) exercises at school, so it would be good for them to have that on that screen, so that the kids could join in with it	Mother of 7-year-old female
6.2	Intervention dosage	
	I think that's good that it does limit that though because I think otherwise, I think doing it once a day is enough	Mother of 4-year-old male
1	*Our aim is we do it twice a week because I do feel that it is really helping*	Mother of 4-year-old male

	Three times a week, fine. I think when it got to the last week, I think they had to do it nearly every day	Mother of 4-year-old male
	I do think your three times, it's like anything when they say at school about reading and stuff, three times a week is probably manageable, especially if you have got other commitments.	Mother of 4-year-old male
	I don't know how many times do you recommend kids do the exercises per day? We were doing it once at home and once at school.	Father of 6-year-old male
2	*It would be a good way of looking back again and seeing how when he was been able to complete it. Because there were times when he felt like he couldn't use it, like, Mum, I need to rest*	Mother of 5-year-old male
	Yeah, she was using it every single day. Weren't you, you were literally on it every day	Mother of 7-year-old female
	Thomas, you've started off well, you were doing it more consistent, but it's kind of, coincided with him feeling a lot better. So, it was asking him to come out of the garden now, stop playing football, which you're not meant to do for three years, and come and do your app, and he was like, oh I want to play football, and then it's like, eight, nine o'clock at night	Father of 7-year-old male

Appendix Z – Thematic table for focus group

Code \ Participant	Child 1 L2	Parent 1 L2	Child 2 S3	Parent 2 S3	Parent 3 S4	Child 4 AH3	Parent 4 AH3	Child 5 L7	Parent 5 L7
1.1 Problems with use		2		2			3		
1.2 Elements increasing use				3		1	1	1	1
1.3 Demonstration of independence		4	1	5		1	2	1	2
2.1 Impact of rewards on use	1	2		2		1	4	1	3
3.1 Positive elements of the app	7	1	4	5	5	2	5	1	4
3.2 Less positive elements of the app		2	2	5	1		1		
3.3 Progression throughout app-testing period		3					1		
4.1 Relationship to current care			1	2			1		1
4.2 Differences to previous care				3	1		1		
4.3 Relevance to stage of Perthes' Disease		2			4		2		
5.1 Changes to functionality of the app		3		2	1		1		
5.2 Ideas for the app	4	5	2	5	4	1	4	1	4
6.1 Educating others				1			1		2
6.2 Intervention dosage		4		1	2		1		1