Timing of surgical intervention for developmental dysplasia of the hip: a randomised controlled trial (Hip ’Op)

Charlotte L Williams, Susie Weller, Lisa Roberts, Isabel Reading, Andrew Cook, Louisa Little, Wendy Wood, Louise Stanton, Andreas Roposch and Nicholas MP Clarke
Timing of surgical intervention for developmental dysplasia of the hip: a randomised controlled trial (Hip ‘Op)

Charlotte L Williams,1 Susie Weller,2 Lisa Roberts,2 Isabel Reading,3 Andrew Cook,4* Louisa Little,1 Wendy Wood,1 Louise Stanton,1 Andreas Roposch5 and Nicholas MP Clarke6

1Southampton Clinical Trials Unit, School of Medicine, University of Southampton, Southampton, UK
2Faculty of Health Sciences, University of Southampton, Southampton, UK
3Faculty of Medicine, University of Southampton, Southampton, UK
4Wessex Institute, University of Southampton, Southampton, UK
5Department of Orthopaedic Surgery, Great Ormond Street Hospital for Children, Institute of Child Health, University College London, London, UK
6Paediatric Orthopaedics, University Hospital Southampton NHS Foundation Trust, Southampton, UK

*Corresponding author

Declared competing interests of authors: Andrew Cook is the vice chairperson of the National Institute for Health and Care Excellence’s Interventional Procedures Advisory committee. It is possible that the committee will issue guidance related to the management of hip dysplasia and, if so, it would use information in this report. Andrew Cook also reports that he is part of a secretariat for a number of National Institute for Health Research (NIHR) committees: in the NIHR Health Technology Assessment programme – the Intervention Procedures Topic Identification, Development and Evaluation (TIDE) panel and the Prioritisation Group; in the NIHR Public Health Research programme – the Research Funding Board and the Prioritisation Group. He is a voting member of the West Midlands Regional Advisory Committee for the NIHR Research for Patient Benefit programme.

Published October 2017
DOI: 10.3310/hta21630

This report should be referenced as follows:

Health Technology Assessment is indexed and abstracted in Index Medicus/MEDLINE, Excerpta Medica/EMBASE, Science Citation Index Expanded (SciSearch®) and Current Contents®/Clinical Medicine.
Criteria for inclusion in the Health Technology Assessment journal

Reports are published in Health Technology Assessment (HTA) if (1) they have resulted from work for the HTA programme, and (2) they are of a sufficiently high scientific quality as assessed by the reviewers and editors.

Reviews in Health Technology Assessment are termed ‘systematic’ when the account of the search appraisal and synthesis methods (to minimise biases and random errors) would, in theory, permit the replication of the review by others.

HTA programme

The HTA programme, part of the National Institute for Health Research (NIHR), was set up in 1993. It produces high-quality research information on the effectiveness, costs and broader impact of health technologies for those who use, manage and provide care in the NHS. ‘Health technologies’ are broadly defined as all interventions used to promote health, prevent and treat disease, and improve rehabilitation and long-term care.

The journal is indexed in NHS Evidence via its abstracts included in MEDLINE and its Technology Assessment Reports inform National Institute for Health and Care Excellence (NICE) guidance. HTA research is also an important source of evidence for National Screening Committee (NSC) policy decisions.

For more information about the HTA programme please visit the website: http://www.nets.nihr.ac.uk/programmes/hta

This report

The research reported in this issue of the journal was commissioned and funded by the HTA programme on behalf of NICE as project number 11/146/01. The protocol was agreed in April 2014. The assessment report began editorial review in November 2016 and was accepted for publication in June 2017. The authors have been wholly responsible for all data collection, analysis and interpretation, and for writing up their work. The HTA editors and publisher have tried to ensure the accuracy of the authors’ report and would like to thank the reviewers for their constructive comments on the draft document. However, they do not accept liability for damages or losses arising from material published in this report.

This report presents independent research funded by the National Institute for Health Research (NIHR). The views and opinions expressed by authors in this publication are those of the authors and do not necessarily reflect those of the NHS, the NIHR, NETSCC, the HTA programme or the Department of Health. If there are verbatim quotations included in this publication the views and opinions expressed by the interviewees are those of the interviewees and do not necessarily reflect those of the authors, those of the NHS, the NIHR, NETSCC, the HTA programme or the Department of Health.

© Queen’s Printer and Controller of HMSO 2017. This work was produced by Williams et al. under the terms of a commissioning contract issued by the Secretary of State for Health. This issue may be freely reproduced for the purposes of private research and study and extracts (or indeed, the full report) may be included in professional journals provided that suitable acknowledgement is made and the reproduction is not associated with any form of advertising. Applications for commercial reproduction should be addressed to: NIHR Journals Library, National Institute for Health Research, Evaluation, Trials and Studies Coordinating Centre, Alpha House, University of Southampton Science Park, Southampton SO16 7NS, UK.

Published by the NIHR Journals Library (www.journalslibrary.nihr.ac.uk), produced by Prepress Projects Ltd, Perth, Scotland (www.prepress-projects.co.uk).
Health Technology Assessment Editor-in-Chief

Professor Hywel Williams  Director, HTA Programme, UK and Foundation Professor and Co-Director of the Centre of Evidence-Based Dermatology, University of Nottingham, UK

NIHR Journals Library Editor-in-Chief

Professor Tom Walley  Director, NIHR Evaluation, Trials and Studies and Director of the EME Programme, UK

NIHR Journals Library Editors

Professor Ken Stein  Chair of HTA and EME Editorial Board and Professor of Public Health, University of Exeter Medical School, UK

Professor Andrée Le May  Chair of NIHR Journals Library Editorial Group (HS&DR, PGfAR, PHR journals)

Dr Martin Ashton-Key  Consultant in Public Health Medicine/Consultant Advisor, NETSCC, UK

Professor Matthias Beck  Chair in Public Sector Management and Subject Leader (Management Group), Queen's University Management School, Queen's University Belfast, UK

Dr Tessa Crilly  Director, Crystal Blue Consulting Ltd, UK

Dr Eugenia Cronin  Senior Scientific Advisor, Wessex Institute, UK

Dr Peter Davidson  Director of the NIHR Dissemination Centre, University of Southampton, UK

Ms Tara Lamont  Scientific Advisor, NETSCC, UK

Dr Catriona McDaid  Senior Research Fellow, York Trials Unit, Department of Health Sciences, University of York, UK

Professor William McGuire  Professor of Child Health, Hull York Medical School, University of York, UK

Professor Geoffrey Meads  Professor of Wellbeing Research, University of Winchester, UK

Professor John Norrie  Chair in Medical Statistics, University of Edinburgh, UK

Professor John Powell  Consultant Clinical Adviser, National Institute for Health and Care Excellence (NICE), UK

Professor James Raftery  Professor of Health Technology Assessment, Wessex Institute, Faculty of Medicine, University of Southampton, UK

Dr Rob Riemsma  Reviews Manager, Kleijnen Systematic Reviews Ltd, UK

Professor Helen Roberts  Professor of Child Health Research, UCL Institute of Child Health, UK

Professor Jonathan Ross  Professor of Sexual Health and HIV, University Hospital Birmingham, UK

Professor Helen Snooks  Professor of Health Services Research, Institute of Life Science, College of Medicine, Swansea University, UK

Professor Jim Thornton  Professor of Obstetrics and Gynaecology, Faculty of Medicine and Health Sciences, University of Nottingham, UK

Professor Martin Underwood  Director, Warwick Clinical Trials Unit, Warwick Medical School, University of Warwick, UK

Please visit the website for a list of members of the NIHR Journals Library Board: www.journalslibrary.nihr.ac.uk/about/editors

Editorial contact: journals.library@nihr.ac.uk

NIHR Journals Library  www.journalslibrary.nihr.ac.uk
Abstract

Timing of surgical intervention for developmental dysplasia of the hip: a randomised controlled trial (Hip ‘Op)

Charlotte L Williams,1 Susie Weller,2 Lisa Roberts,2 Isabel Reading,3 Andrew Cook,4* Louisa Little,1 Wendy Wood,1 Louise Stanton,1 Andreas Roposch5 and Nicholas MP Clarke6

1Southampton Clinical Trials Unit, School of Medicine, University of Southampton, Southampton, UK
2Faculty of Health Sciences, University of Southampton, Southampton, UK
3Faculty of Medicine, University of Southampton, Southampton, UK
4Wessex Institute, University of Southampton, Southampton, UK
5Department of Orthopaedic Surgery, Great Ormond Street Hospital for Children, Institute of Child Health, University College London, London, UK
6Paediatric Orthopaedics, University Hospital Southampton NHS Foundation Trust, Southampton, UK

*Corresponding author andrewc@soton.ac.uk

Background: Developmental dysplasia of the hip (DDH) is a very common congenital disorder, and late-presenting cases often require surgical treatment. Surgical reduction of the hip may be complicated by avascular necrosis (AVN), which occurs as a result of interruption to the femoral head blood supply during treatment and can result in long-term problems. Some surgeons delay surgical treatment until the ossific nucleus (ON) has developed, whereas others believe that the earlier the reduction is performed, the better the result. Currently there is no definitive evidence to support either strategy.

Objectives: To determine, in children aged 12 weeks to 13 months, whether or not delayed surgical treatment of a congenitally dislocated hip reduces the incidence of AVN at 5 years of age. The main clinical outcome measures were incidence of AVN and the need for a secondary surgical procedure during 5 years’ follow-up. In addition, to perform (1) a qualitative evaluation of the adopted strategy and (2) a health economic analysis based on NHS and societal costs.

Design: Phase III, unmasked, randomised controlled trial with qualitative and health economics analyses. Participants were randomised 1 : 1 to undergo either early or delayed surgery.

Setting: Paediatric orthopaedic surgical centres in the UK.

Participants: Children aged 12 weeks to 13 months with DDH, either newly diagnosed or following failed splintage, and who required surgery. We had a target recruitment of 636 children.

Interventions: Surgical reduction of the hip performed as per the timing allocated at randomisation.

Main outcome measures: Primary outcome – incidence of AVN at 5 years of age (according to the Kalamchi and MacEwen classification). Secondary outcomes – need for secondary surgery, presence or absence of the ON at the time of primary treatment, quality of life for the main carer and child, and a health economics and qualitative analysis.
Results: The trial closed early after reaching < 5% of the recruitment target. Fourteen patients were randomised to early treatment and 15 to delayed treatment. Implementation of rescue strategies did not improve recruitment. No primary outcome data were collected, and no meaningful conclusions could be made from the small number of non-qualitative secondary outcome data. The qualitative work generated rich data around three key themes: (1) access to, and experiences of, primary and secondary care; (2) the impact of surgery on family life; and (3) participants’ experiences of being in the trial.

Limitations: Overoptimistic estimates of numbers of eligible patients seen at recruiting centres during the planning of the trial, as well as an overestimation of the recruitment rate, may have also contributed to unrealistic expectations on achievable patient numbers.

Future work: There may be scope for investigation using routinely available data.

Conclusions: Hip ‘Op has highlighted the importance of accurate advance information on numbers of available eligible patients, as well as support from all participating investigators when conducting surgical research. Despite substantial consultation with parents of children in the planning stage, the level of non-participation experienced during recruitment was much higher than anticipated. The qualitative work has emphasised the need for appropriate advice and robust support for parents regarding the ‘real-life’ aspects of managing children with DDH.

Trial registration: Current Controlled Trials ISRCTN76958754.

Funding: This project was funded by the National Institute for Health Research (NIHR) Health Technology Assessment programme and will be published in full in Health Technology Assessment; Vol. 21, No. 63. See the NIHR Journals Library website for further project information.
List of tables

TABLE 1 Screening data by centre 15
TABLE 2 Number of patients recruited by month and centre 17
TABLE 3 Centres in which visits were successfully conducted 18
TABLE 4 Randomisation by treatment arm and stratification factors 21
TABLE 5 Participant DDH diagnosis information 22
TABLE 6 Ossific nucleus presence at time of surgery 23
TABLE 7 Summary of AEs and SAEs 23
TABLE 8 Key reasons preventing the completion of parent interviews 24
TABLE 9 Completed interviews: sample characteristics (n = 14) 25
TABLE 10 Changes to protocol 83
List of figures

**FIGURE 1** Hip ‘Op trial flow diagram  

**FIGURE 2** A CONSORT diagram showing patient screening and recruitment information  

**FIGURE 3** Predicted and actual recruitment
Glossary

**Acetabuloplasty** Reshaping the acetabulum.

**Acetabulum** The ‘socket’ of the ‘ball and socket’ hip joint.

**Anastomotic circulation** Many blood vessels joined together, giving multiple routes for blood to flow. Blocking or damaging one vessel does not prevent blood reaching tissues. Compare with *Terminal circulation*.

**Avascular necrosis** Death of bone components as a result of an interruption of the blood supply.

**Bony epiphysis** An epiphysis in which the cartilage has been replaced by bone.

**Bootstrap** The process of estimating properties of an estimator by repeatedly sampling, with replacement.

**Breech position** Position of a fetus whose bottom, rather than head, is towards the uterus.

**Capsulorrhaphy** Repair of a tear or incision in a joint capsule.

**Chief investigator** The named chief investigator who takes overall responsibility for the conduct of research.

**Chondroepiphysis** An epiphysis that still contains cartilage.

**CONSORT (Consolidated Standards of Reporting Trials) diagram** A diagram that shows the flow of patients through a clinical trial.

**Developmental dysplasia of the hip** A congenital deformation or misalignment of the hip joint.

**Electronic case report form** A tool used to collect data on patients in clinical trials.

**Epiphysis** The end part of a long bone that is, at first, separated from the main part by cartilage, but later fuses with it by ossification.

**Estimator** A rule for calculating an estimate of a given quantity based on observed data.

**Hip arthrography** Radiographical imaging of the hip.

**Iatrogenic** Resulting from the actions of a health-care provider or institution.

**Intermediate profession** Part of the UK National Statistics Socio-economic Classification. These are occupations described as involving clerical work, sales and service.

**Irreducible** Of a dislocated hip, being unable to reseat the hip correctly in its socket.

**Kalamchi and MacEwen grading** A scale for assessing avascular necrosis of the femoral head, ranging from 4 (worst, total damage) to 1 (minimal, changes confined to ossific nucleus).

**Markov model** A stochastic model used to model randomly changing systems in which it is assumed that future states depend only on the present state and not on the sequence of events that preceded it.
Ossific nucleus  The centre of bony transformation in an epiphysis.

Pavlik harness  A brace used for management of developmental dysplasia of the hip. Fabric straps fit around a baby’s chest, shoulders and legs; these hold the legs in a spread position with hips bent, keeping the head of the femur in the acetabulum.

Principal investigator  The lead investigator at a clinical site.

Qualitative  A broad selection of research methods, aimed at exploring the how and why of a process, not just countable outcomes. Compare with Quantitative.

Quality-adjusted life-year  A way of accounting for both the duration and quality of an outcome.

Quanti-qualitative appointment timing  An approach to improving recruitment to randomised controlled trials.

Quantitative  The investigation of a research question using statistical, mathematical or computational techniques. Compare with Qualitative.

Randomised controlled trial  An experiment in which participants are allocated to one of a selection of interventions in a random way, with the aim of reducing or removing bias and confounding.

Spica cast  A shell made of plaster or fibreglass, designed to immobilise one or more limbs.

Subluxed  An incomplete or partial dislocation of a joint.

Tenotomy  Surgical division of a tendon.

Teratologic  Congenital deformity.

Terminal circulation  There is only one route for blood to reach tissues. Blocking the vessel will produce ischaemia and necrosis in distal tissues. Compare with Anastomotic circulation.

Within trial  All the data used for analysis is collected during the study. No estimation is made of what may happen once the study has finished.
# List of abbreviations

<table>
<thead>
<tr>
<th>Abbreviation</th>
<th>Definition</th>
</tr>
</thead>
<tbody>
<tr>
<td>AE</td>
<td>adverse event</td>
</tr>
<tr>
<td>AVN</td>
<td>avascular necrosis</td>
</tr>
<tr>
<td>CarerQol</td>
<td>Care-Related Quality of Life questionnaire</td>
</tr>
<tr>
<td>CI</td>
<td>chief investigator</td>
</tr>
<tr>
<td>CONSORT</td>
<td>Consolidated Standards of Reporting Trials</td>
</tr>
<tr>
<td>DDH</td>
<td>developmental dysplasia of the hip</td>
</tr>
<tr>
<td>DMEC</td>
<td>Data Monitoring and Ethics Committee</td>
</tr>
<tr>
<td>DVD</td>
<td>digital versatile disc</td>
</tr>
<tr>
<td>GP</td>
<td>general practitioner</td>
</tr>
<tr>
<td>HTA</td>
<td>Health Technology Assessment</td>
</tr>
<tr>
<td>HUI-3</td>
<td>Health Utility Index-3</td>
</tr>
<tr>
<td>ON</td>
<td>ossific nucleus</td>
</tr>
<tr>
<td>PedsQL</td>
<td>Paediatric Quality of Life inventory</td>
</tr>
<tr>
<td>PI</td>
<td>principal investigator</td>
</tr>
<tr>
<td>PPI</td>
<td>patient and public involvement</td>
</tr>
<tr>
<td>Q-QAT</td>
<td>quanti-qualitative appointment timing</td>
</tr>
<tr>
<td>QALY</td>
<td>quality-adjusted life-year</td>
</tr>
<tr>
<td>RNOH</td>
<td>Royal National Orthopaedic Hospital</td>
</tr>
<tr>
<td>SAE</td>
<td>serious adverse event</td>
</tr>
<tr>
<td>SCTU</td>
<td>Southampton Clinical Trials Unit</td>
</tr>
<tr>
<td>SD</td>
<td>standard deviation</td>
</tr>
<tr>
<td>TMG</td>
<td>Trial Management Group</td>
</tr>
</tbody>
</table>
Developmental dysplasia of the hip is a common birth problem caused by irregular hip development in babies. Babies born bottom first, and those with a family history of hip problems, are most at risk. When the diagnosis is made at > 3 months of age, surgery is almost always needed. The aim of surgery is to correct the hip position and restore normal movement.

Surgery can be complicated by avascular necrosis, in which the blood supply to the hip is interrupted. This can be devastating for the growing hip, leading to joint damage and, ultimately, hip replacement.

Some surgeons accept that babies treated early may need to be in plaster for longer, but may achieve better results, although there is a greater need for further surgery. Meanwhile, other surgeons believe that intentionally delaying treatment, until the development of a bony ossific nucleus in the hip, may necessitate a bigger operation initially, but will result in less additional surgery in later life.

There is no international agreement among paediatric surgeons regarding whether early or delayed treatment is best. This study was designed to address this question.

This was an ambitious randomised clinical trial that required 636 babies to be recruited and randomised to either early or intentionally delayed surgical treatment and then followed up over 5 years across 15 UK centres. As a precaution, it was decided to have an 18-month run-in period to see if it was likely that this recruitment could be achieved.

The trial closed early as a result of poor recruitment, and so the question could not be answered. Nevertheless, part of the research involved interviews with 14 families and highlighted rich data about getting access to expert orthopaedic care, the impact of the child’s surgery on family life and also what it was like to take part in this trial.
Scientific summary

Background

Developmental dysplasia of the hip (DDH) is one of the most common congenital disorders. Late-presenting cases are synonymous with the need for surgical intervention. Surgical reduction of the hip may be complicated by the development of avascular necrosis (AVN), which can result in long-term problems such as leg length discrepancy and the need for early hip replacement. AVN is an iatrogenic phenomenon that occurs as a result of an interruption to the femoral head blood supply during treatment. Some surgeons delay surgical treatment until the bony ossific nucleus (ON) has developed because this may provide some mechanical protection to the femoral head blood supply, and may thus reduce the chance of AVN developing. However, others believe that the earlier the reduction is performed, the better the result (providing AVN is avoided). Currently, there is no definitive evidence to support either strategy.

Objectives

To determine, in children aged 12 weeks to 13 months, whether or not delayed surgical treatment of a dislocated hip reduces the incidence of AVN at 5 years of age. The main clinical outcome measures were incidence of AVN and the need for a secondary surgical procedure during 5 years’ follow-up. In addition, to (1) qualitatively evaluate parental satisfaction with the strategies and (2) assess NHS and societal costs with the aim of undertaking a health economic analysis.

Methods

Study design

This was a Phase III randomised controlled trial incorporating an internal pilot, and qualitative and health economics analyses. Participants were randomised with a 1 : 1 allocation ratio to undergo early or intentionally delayed surgery for their dislocated hip. The study aimed to recruit 636 children over 4 years; this target recruitment was considered feasible if, within the 18-month internal pilot, 120 children had been recruited. In addition to the internal pilot, a closedown plan had been pre-agreed with the funder, which was activated when the success criteria for the pilot phase of the study were not met.

Settings and participants

Participants were recruited to the Hip ‘Op study from 15 paediatric orthopaedic centres in the UK. Children aged 12 weeks to 13 months with DDH, either having been newly diagnosed or had failed splintage, and who required surgery.

Interventions

Participants were randomised to:

- arm A – early treatment
- arm B – (intentionally) delayed treatment.

The actual procedures carried out were decided by the treating clinician, not by the randomisation or the study protocol.
Outcomes
The primary outcome was incidence of AVN at 5 years of age, as classified according to the Kalamchi and MacEwen grading.

Secondary outcomes were the need for secondary surgery on the affected hip, presence of the ON at the time of primary treatment for dysplasia, quality of life for the main carer and child, a health economic evaluation and the qualitative analysis.

Sample size
Allowing for 10% dropout during the 5-year follow-up period, the total number of patients required was 636 (318 per treatment arm). This sample size had 90% power to detect a 10-percentage point difference between treatment arms (10% vs. 20% AVN rate at 5 years) in a 5% two-sided test.

Randomisation and blinding
Randomisation was via the web-based system TENALEA (TENALEA Randomisation System version 3.0; FormsVision BV, Abcoude, the Netherlands). Allocations were assigned in a 1 : 1 ratio and were stratified by failed splintage and age at diagnosis. Randomisation was carried out once eligibility was confirmed and written consent had been provided. Neither parents nor investigators/surgeons were blinded to the treatment allocation.

Analysis methods: statistical and qualitative
Given the small sample size and early cessation of the trial, it was not possible to perform any of the originally planned analyses for the main trial or health economic aspects. The intention for the main trial was to analyse the presence of AVN by logistic regression, with centre as a random effect and the randomisation stratification factors as fixed effects, using the intention-to-treat population. Secondary analyses were intended to explore the need for further surgery defined according to radiographic findings, and grading of AVN between the treatment arms.

The intention for the health economic analyses was to conduct an analysis of the cost and cost-effectiveness of early versus delayed treatment for infants with DDH. Cost and cost-effectiveness for the ‘within-trial’ period (5 years), and over the expected lifetime of the participant, would have been estimated.

The telephone interviews were audio-recorded, transcribed verbatim, anonymised and analysed thematically using a framework approach. This allowed the interpretation of key issues faced by participants across the sample. Had the trial continued, diachronic case analysis, tracking participants’ accounts over time, would have been used to gain a longitudinal perspective.

Follow-up
It was intended that participants would be followed up at 6 weeks, and 3, 6, 9 and 12 months post surgery, then at 2, 3, 4 and 5 years of age in order to carry out an economic evaluation. It was intended that data be collected between visits by site staff from patient medical records; secondary care resource use data immediately before surgery and at every visit except at 4 months post surgery; and radiography be performed at 5 years of age. For the qualitative work, between 3 and 4 months post surgery, all parents/carers of the participants were invited to complete a demographic questionnaire. Those indicating that they would be prepared to take part in an interview were contacted between 4 and 6 months post surgery and invited to take part in a telephone discussion. The intention was to conduct follow-up interviews when the child reached the age of 5 years.
Results

Patient screening
A total of 118 patients were considered for inclusion. Of these, 44 were not eligible, mainly because of an already existing ON. Of the 74 eligible patients, 44 were not randomised. The most common reasons were that the parent/guardian did not want treatment decided by randomisation and the family did not want to take part in research. The number of children screened varied greatly between centres, and there is no clear relationship between this and the length of time each centre was open to recruitment.

Recruitment
Thirty participants were randomised in the 16 months that the trial was open to recruitment. This represents just over 25% of all children screened, and just over 40% of those who were eligible. There was no relationship between the number of sites open and overall monthly recruitment (median two participants per month). Eight of the 15 sites did not recruit any participants.

Assessing barriers to recruitment and actions taken to increase enrolment
In March 2015, the Trial Management Group discussed the emerging recruitment issue. It was noted that there were fewer than expected entries on screening logs and that eligible patients were refusing the trial. Following this, steps were taken over the remainder of 2015 to investigate the issues and stimulate recruitment.

Centres reported that many children were successfully treated in harness or already had an ON, and that many families with eligible children declined to enter the trial mainly for reasons relating to perceived issues if randomised to the delayed treatment arm. When offered training in methods for study introduction, most centres declined and reported that they did not require any training. Arrangements for a training meeting were abandoned because of a lack of response/interest from site teams. Instead, individual site visits were conducted: during these visits, sites reiterated the lack of eligible children and confirmed that they did not feel any training was required.

Withdrawals
Two of the 30 randomised patients were withdrawn from the study. One family, after having agreed to their child taking part in the study, sought a second opinion, following which surgery was conducted in a different centre and not according to allocation. A second child was the subject of a serious breach (unrelated to patient safety). This incident was fully investigated by the sponsor. The child was immediately withdrawn from the study.

Patient follow-up
Of the 14 patients randomised to early treatment, all underwent surgery during the running of the trial and were followed up to at least 3 months post surgery. Of the 15 patients randomised to intentionally delayed treatment, eight were known to have undergone surgery by the trial end.

Baseline data and demographic characteristics
The majority of patients were aged ≤10 months when they were recruited to the study, and two-thirds had been treated with a splint before presenting for surgery. More girls than boys were recruited to the study, reflecting the prevalence of DDH. The left hip was more commonly affected. The most commonly used diagnostic imaging technique was ultrasound. The median age at diagnosis was 3 months.

Primary outcome
No primary outcome data were collected by trial closure.

Secondary outcomes
Some secondary outcome data were collected by the time of trial closure, including presence of the ON at the time of the primary treatment for dysplasia and some information on surgery outcome. However, no conclusions can be drawn from these minimal findings.
Discussion

Main findings

Study conduct
With a few notable exceptions, most centres cited the lack of suitable patients as the reason for poor recruitment. Initial estimates of patient numbers seen at each centre may have included patients who would, in the study setting, be excluded because they already had an ON. Numbers provided at feasibility were much higher than actual numbers screened. Some sites suggested that this was as a result of an improvement in early detection and with patients being referred at a younger age to specialist centres who have a higher success rate using splints, thereby leaving fewer children needing surgery. Some centres were screening and recruiting as expected, whereas others were not and there was no clear reason why; many centres appeared unenthusiastic or uncommunicative when efforts were made to improve recruitment. Consequently, we conclude that some investigators had difficulties with surgical equipoise and, thus, did not screen or recruit many, or indeed any, patients. The findings from the Hip ‘Op study suggest that it has suffered from the same difficulties as many other surgical trials – lack of robust and honest feasibility, lack of real commitment on the part of some local investigators and their teams and, primarily, lack of surgical equipoise, which was of paramount importance to a study such as Hip ‘Op.

It could be suggested that our initial assumption, that half of all eligible patients would enter the trial, was overly optimistic. We considered it possible – children with DDH in the UK are seen in a comparatively small number of specialist centres, so only a limited number of clinicians had to be engaged in recruitment. In actuality, we managed to recruit 40% of eligible screened patients. The more significant issue would appear to be that fewer patients were screened than was assumed when the study was planned. This might result from a lack of enthusiasm in recruiting centres or from a change in management leading to more DDH being detected and treated before 3 months of age.

Study results
As a result of the early closure of the trial, no primary outcome data were collected, and no meaningful analysis or conclusions could be made from the very small number of secondary outcome data that were collected. In terms of safety, it is worth noting that no significant adverse events occurred: this was as expected because all procedures within the trial were as per standard practice.

Qualitative aspects
The qualitative data generated rich data around three key themes: (1) access to, and experiences of, primary and secondary care (including challenges of raising concerns); (2) the impact, and burden, of surgery on family life (including financial impact and implications for parental physical/mental well-being); and (3) participants’ experiences of being in the trial. These findings have relevance for both clinicians and researchers in developmental dysplasia.

Methodologically, this pilot work, if extended further, could contribute to the growth and application of qualitative work within clinical literature, particularly paediatric orthopaedics, in which DDH is an important area of interest and qualitative research is underutilised.

Strengths and limitations
The main strengths of Hip ‘Op were (1) the pragmatic design, (2) the study management, (3) the 18-month pilot (it established a clear cut-off point that prevented wasting resources on a failing trial) and (4) inclusion of the qualitative aspect. The major limitation is that the study closed without recruiting a sufficient number of patients to answer the trial question.
Lessons for the future
If we were setting up Hip Op again, ideally we would conduct face-to-face interviews with the team at each prospective site to investigate potential pitfalls, loopholes and concerns at the outset. Second, prior to opening, we would agree with the sites a standardised way in which to present the trial in an unbiased manner to minimise numbers who decline to take part. We would undertake more intensive work with patient support organisations to bring the study to the attention of parents before their first orthopaedic outpatient attendance. We would also investigate whether or not it was feasible to conduct the study in centres internationally rather than in the UK only. We would also investigate expected eligible patient numbers at each site more closely.

Future research
The question posed by Hip Op is still valid and remains unanswered; however, it is clear that it cannot currently be answered in the UK with a randomised trial. Similar research is under way internationally, and it is likely that data from Hip Op could contribute to a meta-analysis from this effort. The qualitative part of the study could have far-reaching impacts on clinical decision-making and practice, and family support. This study has identified areas where information could be improved for families of children who are diagnosed with DDH and require surgery, and further funding is being sought to explore the experiences of a greater diversity of families and to examine the long-term impacts. In addition, the possibility of at least partially addressing this question using routinely collected data is being explored.

Conclusion
Hip Op has highlighted how important accurate feasibility information up front, as well as commitment from all participating investigators, is when conducting surgical research, and how lack of these important elements can lead to a spectacular inability to recruit. The Hip Op trial was novel because of the inclusion of the qualitative research aspects. The study has underlined how important these results are, not only in terms of patient participation in clinical research, but in in terms of highlighting the need for appropriate advice and robust support for parents regarding the ‘real-life’ aspects of managing an infant with DDH.

Trial registration
This trial is registered as ISRCTN76958754.

Funding
Funding for this study was provided by the Health Technology Assessment programme of the National Institute for Health Research.
Chapter 1 Introduction

Background and rationale

In the UK, hip instability at birth occurs with an incidence of 15–20 per 1000 live births. In many cases the instability resolves spontaneously. In 7–15 per 1000 live births, some form of active management is required, usually initially with a splint or harness. Early treatment with a splint is effective in 85% of cases, if treatment commences in the first 6–8 weeks of life. However, despite clinical and ultrasound screening programmes, late-presenting cases (aged > 3 months) persist. Such cases are synonymous with the need for surgery, but it is unclear whether it is better to undertake that surgery promptly or after a delay of possibly several months.

When surgery is indicated, then the surgeon may undertake either an open (formal surgical) reduction or a closed reduction after adductor tenotomy. The goal is for the femoral head to sit concentrically with the acetabulum. Reduction is confirmed as concentric by hip arthrography. Both types of intervention may be complicated by the development of avascular necrosis (AVN), which occurs as a consequence of partial, temporary or complete interruption of the blood supply to the femoral head and is entirely iatrogenic.

Before 8–10 months of age, the femoral head is a chondroepiphysis and the blood supply is endarteriolar. With the development of the bony epiphysis [which may be delayed in developmental dysplasia of the hip (DDH)], the blood supply becomes anastomotic. It has been hypothesised that the anastomotic circulation renders the femoral head less vulnerable to compression and, therefore, vascular injury. Accordingly, some surgeons delay surgical intervention until after the bony epiphysis has appeared, which can be identified by ultrasound. However, delay also allows the dysplasia to progress and, therefore, surgery is not usually intentionally delayed beyond the age of 12 or 13 months, although by this time an epiphysis will have appeared in most cases.

The incidence of AVN is variously reported as occurring in 10–50% of cases of DDH and adversely affects outcome because of proximal femoral deformity, eccentric growth, poor femoral head containment and, consequently, leg length discrepancy. An early closed reduction requires more plaster changes and most cases will require a secondary procedure to address residual acetabular dysplasia. Delayed open reduction is usually definitive treatment because acetabular dysplasia can be addressed at the primary procedure. There is no international consensus and the only meta-analysis carried out was not conclusive in respect of either strategy.

The proposed research aimed to address the clinical effectiveness and cost-effectiveness of intentionally delayed versus early surgical intervention in established DDH.

Rationale

Proponents of intentional delay hypothesise that the appearance of the bony ossific nucleus (ON) within the femoral head confirms mechanical resistance to compression and, hence, reduces the risk of AVN (iatrogenic ischaemic injury). Prior to the appearance of the ON, the chondroepiphysis is more vulnerable and secondary surgical procedures are more likely. It was intended that the cost-effectiveness be interrogated by health economic studies. A preliminary feasibility study was conducted to address stakeholder and consumer willingness to take part, likelihood of recruitment and approximate recruitment rate.
Aims and objectives

The objective of the trial was to determine, in children aged 12 weeks to 13 months, if delayed treatment of a dislocated hip, in the absence of the proximal femoral ON, could reduce the incidence of AVN in children at 5 years of age. The main clinical outcome measures were the incidence of AVN and the need for subsequent secondary surgery during the 5 years’ follow-up. We also sought to (1) qualitatively evaluate parental satisfaction with the adopted strategy and (2) assess NHS and societal costs with the aim of undertaking a health economic analysis.

Assessed health technologies

We aimed to assess the timing of surgical reduction of the hip. The strategies being compared were (1) early surgery soon after diagnosis (before the appearance of the ON) and (2) intentionally delayed surgery (soon after the appearance of the ON). The actual procedure carried out was decided by the treating clinician and was not determined by the randomisation allocation or the study protocol.

This report contains the results of the Hip ‘Op trial. The trial closed early because of poor recruitment, after < 5% of the target recruitment had been reached. Here we describe the valuable insights gained from the qualitative aspect on the trial, as well as highlighting the challenges faced and lessons learnt from the experience.
Chapter 2  Methods

Trial design

The Hip ‘Op trial was a pragmatic, multicentre (UK), Phase III, randomised controlled trial incorporating an internal pilot, and qualitative and health economics analyses. Participants were randomised between early and intentionally delayed surgical reduction of a dislocated hip with a 1 : 1 allocation ratio (Figure 1).

We needed to recruit a total of 636 children aged 12 weeks to 13 months with a dislocated hip, in the absence of the proximal femoral ON (318 per treatment arm). The children were stratified at randomisation by failed splintage and age at diagnosis (≤ 10 months or > 10 months).

During study development, it was recognised that recruitment to this trial would be very challenging. As such, an internal pilot was planned to assess the ability to recruit and likely generalisability of findings. In addition, a closedown plan had been pre-agreed with the funder and was to be activated if, as did

![Hip 'Op trial flow diagram](image-url)

**FIGURE 1** Hip ‘Op trial flow diagram. CT, computerised tomography; GP, general practitioner.
happen, the success criteria for the pilot phase of the study were not met. The internal pilot would have been deemed successful if, during the first 18 months of recruitment:

- at least 10 centres were actively recruiting patients
- at least 120 patients had been recruited
- there had been sufficient mean monthly recruitment (based on the period since the tenth centre started to recruit) to expect to reach 636 patients by the end of month 56.

There was no change to the trial design after commencement of the trial.

Participants

**Eligibility criteria**

Infants were eligible if they satisfied the following criteria:

- were aged 12 weeks to 13 months with either
  - a new diagnosis of DDH
  - failed splintage up to 12 weeks of age
- were born at ≥ 30 weeks’ gestation
- required surgical reduction of the hip (open or closed)
- were fit for surgery – the decision to include in the study was entirely at the discretion of the operating surgeon
- had a parent or guardian willing to give consent to treatment and to complete questionnaires and follow-up.

Infants were not eligible if they satisfied any of the following criteria:

- were aged > 13 months
- had neurological or syndromic teratologic dislocation of the hip – if in doubt, such infants were not included
- were born at < 30 weeks’ gestation
- had had any previous surgical treatment for hip dysplasia (closed reduction, open reduction or any form of tenotomy)
- had existing AVN
- had existing ON.

**Changes to eligibility criteria after commencement of the trial**

Following the first Data Monitoring and Ethics Committee (DMEC) meeting, held in January 2015, an extra exclusion criterion for babies born at < 30 weeks’ gestation was added. This was implemented from May 2015.

**Setting and recruitment pathway**

Participants were recruited to the Hip ‘Op study from paediatric orthopaedic centres in the UK (see Appendix 1). Identification and recruitment of eligible children were dependent on all surgeons within a given centre actively searching for patients and presenting the trial to the families with equipoise (i.e. in a balanced manner).
Individual surgeons could conduct surgery as per the treatment allocation, but did not have to be delegated study research responsibilities (i.e. as long as a treating surgeon agreed to perform the surgery as per the timing allocated at randomisation, that surgeon did not have to be part of the local research team).

Interventions

Trial participants were randomised to:

- arm A – early treatment
- arm B – (intentionally) delayed treatment – surgery to have taken place within 2–4 weeks of the appearance of the ON (unless exceptional circumstances required it to be delayed further).

The actual procedures carried out were decided by the treating clinician and were not determined by the randomisation allocation or the study protocol; however, at the time of writing, there are only a small number of established surgical procedures in use. Only the timing of surgery was determined by randomisation; normal local surgical protocol was followed for the surgery itself. Normal clinical and orthopaedic assessment and care were carried out pre and post surgery.

Monitoring for the appearance of the ossific nucleus

For participants randomised to have delayed treatment, 6-weekly imaging (the type of imaging performed was as per local practice) was considered ideal to monitor for the appearance of the ON and, thus, trigger surgery.

Outcomes

Primary outcome

The primary outcome was to assess the incidence of AVN at 5 years of age with AVN assessment classified radiologically according to the Kalamchi and MacEwen grading (grades I–IV). It was intended that a subset would also be classified by an independent panel of radiologists/surgeons blinded to treatment arm.

Secondary outcomes

Secondary outcomes were:

- the need for secondary surgery on the affected hip (for subluxation/dysplasia/AVN), as recorded from a review of medical records during follow-up visits
- presence or absence of the ON at the time of primary treatment for dysplasia (assessed from radiographs taken within 24 hours of the index reduction)
- quality of life for the main carer [assessed using the Care-Related Quality of Life (CarerQol) questionnaire]
- quality of life for the child [assessed using the Oucher Pain Scale, Paediatric Quality of Life (PedsQL) inventory or the Health Utility Index-3 (HUI-3)]
- health economic evaluation
- qualitative analysis.

Adverse events

Adverse events (AEs) were captured in the purpose-designed electronic case report form. Serious adverse events (SAEs) were reported via the purpose-designed paper SAE report form.
The protocol stated that expected complications of DDH surgery should be reported as AEs and not SAEs. Expected complications were specified in the Hip ‘Op protocol as follows:

- failed location
- wound infection
- redislocation
- unscheduled change of plaster.

**Sample size**

The overall aim of this study was to effect clinical practice change by definitively establishing if one of the two treatment strategies, either early or delayed surgery, is superior in reducing the occurrence of AVN following surgery. In order to effect such a change within the practising surgical community, a 10-percentage point difference in AVN rate between treatment arms was considered by the clinical community to be the minimum important difference. Allowing for 90% power, to detect a clinically meaningful difference between treatment arms in a 5% two-sided test indicated that 286 patients per arm were required. This assumed proportions of AVN, defined as grades II–IV, of 20% (ON absent) versus 10% (ON present) or vice versa (i.e. to detect an odds ratio of 0.444). Allowing for 10% dropout during the 5-year follow-up period, the total number of patients required was 636 (318 per treatment arm). This sample size calculation was performed using nQuery Advisor version 7.0 (Statistical Solutions, Boston, MA, USA), using data from figure 3 from the systematic review by Roposch et al.4

**Reassessment of sample size**

Rates of AVN occurrence are known to vary widely,5 which suggested that the treatment effect could have been considerably larger than 10%. The Hip ‘Op trial, thus, had a re-estimation of the sample size pre-planned for after the 300th patient reached the 6-month post-surgery follow-up (as the majority of AVN occurs within 6 months of surgery). This would have assessed whether or not the sample size for the study needed to change, for example if a bigger effect or a smaller event rate had been observed. The reassessment of sample size was not undertaken because of the premature termination of the trial as a result of under-recruitment.

**Randomisation**

Randomisation was via the independent, web-based system TENALEA. Allocations were assigned in a 1 : 1 ratio and were stratified by failed splintage and age at diagnosis (≤ 10 months or > 10 months). Randomisation was carried out by site staff only once eligibility for the trial was confirmed and the child’s parent/guardian had provided written informed consent. Each participant was automatically assigned a unique participant identification number via the TENALEA system during the randomisation process.

Randomisation of participants via the TENALEA system was restricted to the principal investigator (PI) and appropriately delegated site staff, each of whom had individual password-protected login information. Site staff could perform only randomisation; no other randomisation parameters could be altered by research staff at sites. All site staff with a TENALEA login (at the site of the randomisation), as well as the Southampton Clinical Trials Unit (SCTU) trial team, were automatically sent a randomisation notification within minutes of it taking place.

Eligible premature babies were not randomised until they reached 12 weeks past their estimated due delivery date. However, the actual date of birth of such children was entered into the TENALEA system for randomisation.
Blinding

Owing to the nature of the trial, neither parents nor investigators/surgeons were blinded to the treatment allocation.

It had been planned that the independent panel of assessors be blinded for the evaluation of AVN from the radiographs taken at 5 years of age; however, no participant reached this stage in the trial and, thus, this blinded assessment did not take place.

Data collection

Main trial

All patients had the usual assessments needed as part of their standard treatment as determined by their treating surgeon.

Paper screening logs recording the number of eligible patients seen, numbers randomised and reasons for patients not entering the trial were collected from sites during the trial, when possible.

Given the small sample size and early cessation of the trial, it was possible to collect only a very limited number of the data that had been originally planned. The intended data collection was as follows.

Clinical data from medical records was to be collected for use in the trial and transcribed to the purpose-designed electronic case report form. The following data were required as a minimum.

- Baseline assessments: confirmation of diagnosis and eligibility. In addition, the preoperative radiological grade of dislocation and acetabular index measurements (when assessable from the type of imaging performed).
- During the ‘delay’ period in the late-treatment arm: details of imaging used to determine appearance of the ON (i.e. ultrasound/radiography).
- Treatment: summary details of surgical intervention carried out and any complications.
- Follow-up: details of routine imaging carried out, grading of images, incidence of AVN and summary details of further treatment required.

It was intended that participants would also be followed up at 6 weeks, 3, 6 and 9 months, and 1 year post surgery, and at 2, 3, 4 and 5 years of age in order to carry out an economic evaluation. The exact timing of these visits may have been earlier or later than the times stated above, dependent on local clinical practice. It was not envisaged that extra visits to clinic would be required; however, if necessary, parents may have been asked to bring their child to clinic for an interim visit.

It was intended that:

- data be collected for each interval between visits by site staff from patient medical records
- secondary care resource use data be collected immediately pre surgery and at every visit except at 4 months post surgery
- AEs be collected at all visits after the date of consent
- radiography be performed at 5 years of age in order to assess AVN (which was the primary end point).

An independent panel of assessors would have evaluated, in consensus, the 5-year anteroposterior pelvis radiograph for the presence of AVN using the Kalamchi and MacEwen classification. Electronic copies of these radiographs were to be sent to the SCTU by each hospital. The panel would have evaluated these blinded to intervention, nature of the treatment, site and patient details.
Health economics
Given the small sample size and early cessation of the trial, it was possible to collect only a very limited number of the health economic data that had been originally planned. The intended data collection was as follows.

Health-care resource utilisation was to be measured directly from patient records and diaries completed by the participant’s parent.

Site staff would have collected secondary care resource use data from hospital records, specifically inpatient stays, length of stay (day cases) and reason for admission; accident and emergency department attendances; and outpatient visits, type of visit and reason for visit.

Parental diaries were to be used to collect data on primary care and community care resource use, and costs borne by families, as follows:

- primary care and community care contacts [i.e. general practitioner (GP)/nurse visits at practice or health centre, at home or telephone calls]
- other NHS contacts (i.e. listing the type of contact or the health-care professional contacted, where the contact took place, the date of the contact, whether NHS or private, and also the money spent)
- medications taken, including the name of the medication, the dosage taken each time, the number of doses taken each day, the number of days the medication is taken, whether the medication was prescribed by a doctor or nurse, or bought over the counter, and also the money spent
- participants’ parents were also asked to record if they incurred other expenditures in relation to the child’s hip condition, how they financed their health expenditures, the costs borne by them and their family (including if the condition of their child has affected their work, private and social life) and their child care costs.

This information was to be collected both in the first year post surgery and annually between the ages of 2 and 5 years of the child. It was intended that the diaries be distributed at each visit and collected from the parents at their next clinic visit.

Health-related quality of life was to be measured using the following three validated instruments: CarerQol questionnaire for the parent/caregiver, the Oucher Pain Scale and PedsQL inventory.

The CarerQoL questionnaire measures care-related quality of life in informal caregivers. This instrument combines the information density of a burden instrument (encompassing seven important burden dimensions) with a valuation component (a visual analogue scale for happiness).

The Oucher Pain Scale is a poster-like instrument designed to help children provide self-reports of the intensity of their pain. The six-picture photographic scale was intended to be used in Hip ‘Op.

The PedsQL inventory is a generic health-related quality-of-life measure in children to be used across various paediatric chronic health conditions. The PedsQL Generic Core Scales for the specific age groups from baseline to 5 years of age was to be used in Hip ‘Op. These were designed to measure the core dimensions of health as delineated by the World Health Organization, as well as role (school) functioning: the PedsQL Infant 1–12 months, the PedsQL Infant 13–24 months, the PedsQL Parent Report for Toddlers (aged 2–4 years) and the PedsQL Parent Report for Young Children (aged 5–7 years).

Except for the Oucher Pain Scale, all the instruments would have been completed by parents and collected by site staff from parents at relevant clinic visits, then returned to the SCTU.
Qualitative
As well as considering the outcome from early versus intentionally delayed surgery, the aim of the qualitative study was to explore the experiences of families (families, conceptually broad, is used to move discussion beyond the impact on parents to include siblings, grandparents and other carers) to determine what it is like managing with a child undergoing surgical intervention for DDH. Rigorous qualitative research has the capacity to shed light on the broader implications of treatment and surgery by highlighting the circumstances underlying parents’ experiences of, and responses to, their child’s condition. Essentially, it provides detailed insights into the complexity of experiences, providing answers to ‘how’ and ‘why’ families experience the journey in different ways.

The qualitative element of the trial was intended to be a longitudinal study comprising repeat, in-depth telephone interviews capturing a diversity of parents'/carers’ experiences.

Between 3 and 4 months post surgery, all parents/carers of the participants in the trial were invited by the clinical team to complete a demographic questionnaire that explored, in brief, their motivations for, and experiences of, taking part in the trial as well as background (baseline) information about their family and employment circumstances. This material was intended to inform both the purposive sampling and analysis of the interviews. Those indicating in their questionnaire that they would be prepared to take part in an interview, were contacted between 4 and 6 months post surgery and invited to take part in a telephone discussion. Therefore, no relationship was established prior to the study commencing. The interviewer was introduced to participants as a researcher. Interviews were audio-recorded, transcribed verbatim and analysed thematically using a framework approach and the researcher recorded contextual field notes to aid analysis, reflexivity and purposive sampling.

Telephone interviewing has the advantage that it permits access to a nationally distributed sample, some of whom may be otherwise hard to access; is time and cost-efficient; provides participants with the flexibility to incorporate an interview into family life, the demands on which are likely to be exacerbated by DDH treatment; and has been shown to provide opportunities for less confident participants to speak without the pressure of the presence of a researcher.

In addition to the formal trial consent process, participants were invited to indicate their willingness to take part in a telephone interview as part of their response to the demographic questionnaire. Verbal consent was sought at the beginning of the interview, including permission to record the discussion for the purposes of analysis. All participants were offered the opportunity to ask questions about the intention, process and potential outcomes of the interviews, and were informed that they may pause or terminate the discussion at any point. The topic guide for the semistructured interviews is shown in Appendix 2. At the end of the interview, once participants were fully aware of what they had divulged, consent was reaffirmed to both use the material disclosed and to recontact families when their child reached the age of five for a further interview. The framework data were analysed (by SW and LR) and managed using NVivo (version 10; QSR International, Warrington, UK).

The original aim was to conduct pilot interviews with 20 parents/carers (10 in each of the treatment arms) and, based on these responses, a further 30 would be sampled purposively to include a diversity of family circumstances and experiences for both intervention groups (15 in each). The interviews focused on the family’s DDH journey, including any family history; the process of diagnosis; emotional responses; perceptions of their child’s progress; adjustment to new caring tasks; relational issues and intimacy with their child; effects on parental well-being; implications for family resources, including the cost of hospital visits and additional equipment, and impacts on employment; child care options/constraints; sources/networks of support; and experiences of surgery and aftercare.

The final intention was to conduct follow-up interviews when the child reached the age of 5 years to understand, longitudinally, how parents manage the impact of treatment and rehabilitation. This trial would have generated original qualitative data from 100 interviews, providing a rich data set on patient
experience following DDH surgery. It was intended that the qualitative data be used to help interpret the clinical and quantitative data by considering patient experience alongside outcome, thereby considering the application of early and intentionally delayed surgery in the context of everyday life.

Despite the early cessation of the trial, it was possible to collect a large number of valuable qualitative data for the pilot phase. Of the 20 families needed for the pilot phase, 14 participated in telephone interviews at 4–6 months post surgery, and the data were analysed as far as possible as originally intended.

**Long-term follow-up**

It was intended that parents be asked for permission for their child to be followed up beyond the trial period by collecting routine data from their medical records and Hospital Episode Statistics to determine the need for subsequent intervention (further surgery, hip replacement, diagnosis of arthritis, etc.). Long-term follow-up would have allowed establishment of a cohort to understand the long-term consequences, if any, of the interventions.

Consent for long-term follow-up would have only been sought at approximately 1 year post surgery. Refusal would not have affected participation in the main trial.

**Statistical methods and data analysis**

**Main trial**

Given the small sample size and early cessation of the trial, it was not possible to perform any of the originally planned analyses for the main trial.

The original intention was to analyse the presence of AVN by logistic regression with centre as a random effect and the randomisation stratification factors as fixed effects, thus yielding an odds ratio with 95% confidence intervals for treatment effect, using the intention-to-treat population. Secondary analyses were intended to explore the need for further surgery defined according to radiographic findings, as well as further investigation on the grading of AVN between the treatment arms. In addition, it was the intention to explore the presence or absence of the ON at the time of the primary treatment. A prespecified subgroup analysis would have investigated the effect of failed splintage on the treatment effect (by including a treatment interaction in the regression model), although the study was not powered to detect such an effect.

Instead, analysis has been limited to the presentation of the characteristics of the randomised participants, details of the surgery they underwent, a report of AEs and SAEs, and any follow-up as far as they were known by the trial closure.

**Health economic aspects**

Given the small sample size and early cessation of the trial, it was not possible to perform any of the originally planned analyses for the health economic aspects of the trial. Intended analysis of this part of the trial is as follows.

The intention was to conduct a detailed analysis of the cost and cost-effectiveness of early versus delayed treatment for infants with DDH using the data collected as described. Cost and cost-effectiveness for the ‘within-trial’ period (5 years), and over the expected lifetime of the participant, would have been estimated using accepted economic evaluation methods.

In the primary economic analysis, it was intended that the costs be assessed from the perspective of the NHS and personal social services. Cost components included in the analysis would have consisted of the costs of the intervention by type, diagnostic imaging, secondary operations by type, overall hospital length of stay, outpatient attendances, readmissions, all primary care contacts and all prescribed treatments. Secondary economic analysis would have additionally included monetary costs borne by families.
As stated, the volume of resource use for each cost component would have been measured from NHS electronic records (for secondary care resource use) and from parental diaries and questionnaires (primary care contacts, costs borne by patients and families). Unit costs would have been taken from standard published sources. Unit costs would have been multiplied by mean resource use for each cost component to calculate mean costs per patient in each arm of the trial.

Cost-effectiveness measures at the 5-year end point would have been the incremental cost per AVN averted and the incremental cost per quality-adjusted life-year (QALY) gained. The number of cases of AVN averted would have been based on trial outcomes.

Health-related quality of life and utilities
The intention was to use the HUI-3, collected at the last visit, to derive utilities for children at the age of 5 years. The HUI-3 is valid for children aged ≥5 years and has been used widely in this context.9 Because the HUI-3 has not been validated in children aged <5 years (utility measures for younger children do not exist), the intention was to collect longitudinal data on health outcomes using the PedsQL inventory at baseline, 3, 6, 9 and 12 months, and at 2, 3, 4 and 5 years (PedsQL has been validated for infants from the age of 1 month), and the Oucher Pain Scale in participants who were aged 2, 3, 4 and 5 years (the Oucher Pain Scale has been validated only for children from aged ≥2 years). Such longitudinally collected data would have allowed the determination of the relationship between the HUI-3 and PedsQL inventory/Oucher Pain Scale at 5 years. The intention was to use regression analysis to model the relationship between the HUI-3 (total score and the ambulation, pain, emotion and cognition attributes) using PedsQL inventory/Oucher Pain Scale summary scores as the independent variables at 5 years, assuming the mapping was time invariant, and then the estimated coefficients would have been used to predict the HUI-3 scores from baseline. This would have allowed QALYs to be modelled for study participants for the duration of the trial.

In addition, it was intended to determine the impact of a child’s condition on the caregiver. This would have been done by measuring health-related quality of life of the same parent/carer using the CarerQoL questionnaire.

Cost-effectiveness would have been calculated as the mean cost difference between early versus delayed treatment divided by the mean difference in outcomes (occurrence of AVNs/QALYs). This would have given the incremental cost-effectiveness ratio, and its confidence intervals would have been estimated using bootstrapping techniques of the mean cost and outcomes differences.10 The bootstrap replications would have been used to construct a cost-effectiveness acceptability curve, which would have shown the probability that delayed surgery is cost-effective at 5 years for different values of the NHS’s willingness to pay for an additional QALY. In addition, it was intended that deterministic sensitivity analysis be performed.

In the lifetime analysis, it was intended that cost-effectiveness be calculated in terms of the incremental cost per QALY gained. No previous analyses on the cost-effectiveness of differences in delayed/immediate surgery for DDH exist. It was intended that a de novo cost-effectiveness model based on pre-existing work be developed.11,12 Data from these studies, and data collected in the trial, would have enabled the development of a new model taking into account long-term outcomes (i.e. osteoarthritis of the hip and hip replacement surgery). Two Health Technology Assessment (HTA) programme-funded studies on the cost-effectiveness of hip replacement surgery would have provided further data to develop this model.13,14 Given the clinical nature of DDH (operations may happen more than once, risk of osteoarthritis is continuous over time, timing of events is important), a Markov model would have been constructed. The health states in this model would have reflected the various disease pathways (e.g. primary treatment of DDH, treatment for AVN, treatment for acetabular dysplasia, physical disability, onset of osteoarthritis, death). Following decisions about the model structure, we would have derived a list of parameter estimates required for the model. Finally, it was intended to undertake deterministic and probabilistic sensitivity analysis that would have been used to construct cost-effectiveness acceptability curves.15
Qualitative aspects
The key to qualitative research is to get as broad a sample as possible to maximise transferability and, therefore, the implementation of findings. Fourteen of the 20 originally planned pilot telephone interviews were conducted as part of Hip ‘Op and were analysed as originally intended. The 14 interviews resulted in 722 minutes of audio data. All interviews were audio-recorded, transcribed verbatim, anonymised and analysed thematically using a framework approach. This is a systematic means of analysis, which not only provides in-depth insights but also is founded on developing a robust audit trail, thereby enhancing the credibility of the work. Using a framework agreed by the qualitative investigators, and generated using a priori codes, devised in relation to the project remit, and in vivo codes, based on themes emerging from the data, the transcripts were coded and categorised using the software package NVivo (version 10). Case-by-theme matrices were generated to allow exploration across the cases, generate themes and links between codes, and to explore relationships using the baseline attributes of each participant (e.g. gender, familial circumstances, level of education, employment). This allowed the interpretation of key issues faced by participants across the sample. Had the trial continued, diachronic case analysis, tracking participants’ accounts over time, would have been used to gain a longitudinal perspective. Data from interview participants’ entries in the parental cost diaries have also been drawn on in the analysis of the qualitative data.
Chapter 3  Results

Centres

A total of 15 centres were participating in the Hip ‘Op trial when the study closed. Originally, it was planned that the Hip ‘Op trial would recruit participants from 13 centres in the UK. Of these original 13 centres, 11 received all necessary approvals and opened, and two (Plymouth and Sunderland) withdrew prior to receiving approvals. A further six UK centres subsequently expressed interest in the trial: four of these received all necessary approvals and opened, and the remaining two (Coventry & Warwick and Leicester) were at various stages of the approvals process when the study closed (see Appendix 1). No centres in the devolved nations expressed interest in taking part; thus, all 15 centres that participated in the Hip ‘Op trial were in England. Of the centres that participated, 10 were open within the first 4 months (October 2014–January 2015). The remaining five sites opened subsequently during 2015.

During the recruitment period from October 2014 to January 2016, participants were recruited from seven centres: Southampton, Alder Hey, Nottingham, Newcastle, Oxford, The Royal National Orthopaedic Hospital (RNOH) Stanmore and Great Ormond Street Hospital.

Screened patients

A total of 118 patients were considered for potential inclusion in the trial (see Figure 2). Of these, 44 were not eligible: the main reason for ineligibility (n = 28) was that these children already had an ON. The other main reason for ineligibility (n = 14) was ‘other’, which constituted the following (as assessed from the comments added by site staff): eight resolved, two did not need surgery, two with an unknown reason, one hip was irreducible and one subluxed.

Of the 74 eligible patients, 44 were not randomised to the study. The most common reasons were that the parent/guardian did not want their child’s treatment decided by randomisation (n = 17) and that the family did not want to take part in a research study (n = 9). Interestingly, three of the families that refused to take part in the trial specified that they wanted to have early surgery. Further reasons are detailed in the Consolidated Standards of Reporting Trials (CONSORT) diagram (Figure 2).

Table 1 shows the key screening data broken down by centre. The number of patients screened varied greatly between centres and there is not a clear relationship between this and the length of time each centre was open to recruitment. In addition, there is large variability between centres for the percentage of children screened with an existing ON (range 0–67%) and randomised (range 0–83%). This large variability probably reflects inconsistent screening/screening log completion.

Recruitment

A total of 30 participants were randomised into the study between October 2014 and the end of January 2016, when the study was closed to recruitment by agreement with the study funder. This represents just over 25% of all children screened, and just over 40% of those who were eligible. The pre-agreed 6-month closedown was initiated at the end of January 2016 and all study activities had ceased by the end of July 2016. Centres that had recruited participants were asked to stop collecting data on 9 May 2016, but telephone interviews for the qualitative work continued until the end of June 2016.
Cumulative and predicted recruitment is shown in Figure 3. The actual number of participants recruited began to fall below predicted recruitment in February 2015, approximately 4 months into the recruitment period, despite two-thirds of the sites having already been opened. By the end of January 2016, when the study was closed to recruitment, < 30% of the predicted number of participants to that date had been recruited.

**Figure 2** A CONSORT diagram showing patient screening and recruitment information. ASAP, as soon as possible; LAC, local authority care.
Table 2 shows that a total of 30 participants were randomised to the Hip ‘Op trial in the 16 months that the study was open to recruitment. As can be seen, there was no relationship between the number of sites open and overall monthly recruitment. Total monthly recruitment was quite consistent, ranging between zero and five participants, with a median of two participants per month.

Table 2 shows that recruitment between sites was less consistent, with 8 out of the 15 sites not recruiting any participants at all. Of the seven sites that did recruit, Southampton and Alder Hey recruited the most successfully, with 15 and six participants, respectively. Nottingham (n = 3), RNOH (n = 2), Great Ormond Street Hospital (n = 2), Oxford (n = 1) and Newcastle (n = 1) also recruited participants.

Assessing barriers to recruitment and actions taken to increase enrolment
In March 2015, the Trial Management Group (TMG) discussed the emerging recruitment issue. During this meeting, it was noted that there were fewer than expected entries on screening logs and that eligible patients were refusing the trial. Therefore, it was decided that the chief investigator (CI) should correspond directly with all site PIs to encourage them to record all screening entries, to use the study information film when introducing the study to families and to reiterate the importance of introducing the study with equipoise (i.e. in a balanced manner).
In May 2015, the SCTU e-mailed sites to ask for detailed answers focusing on why so few patients were being screened and what the most successful way to introduce the study to families was. A common report from sites was that many children were successfully treated in harness, and that many families with eligible children declined to enter the trial mainly for reasons relating to perceived practical issues if randomised to the delayed treatment arm (e.g. delayed return to work from maternity leave). Such issues had not been encountered at the CI’s site, Southampton (with an 83% screen-to-enrolment rate); thus, it seemed likely that sites with a poor screen-to-enrolment rate may not have been employing an effective patient information strategy.

The responses to this e-mail were reviewed by the TMG in June 2015, when it was decided that a description of the CI’s successful method should be formally sent to all sites; arrangements should be made for an investigator training meeting; quanti-qualitative appointment timing (Q-QAT) involvement should be investigated.17 In addition, a study-specific website should be built to raise the internet profile of the study, and sites should be provided with desktop reminder cards. In addition, it was decided that a teleconference with site research nurses/physiotherapists/co-ordinators should be arranged to provide training and discuss screening and recruitment issues; this teleconference was held in June 2015 and was well received. The following points arose from discussion.

- Many cases were successfully treated with a Pavlik harness.
- The families were confident in the study only if the PI/surgeon provided the explanation, or at least played the major role in the explanation.
- Some PIs/surgeons did not seem to be using the best way to explain the study and, thus, further structured guidance/training was thought to be necessary.
- Some PIs were not engaging with, and supporting, the local study teams and this had a negative impact on recruitment and morale.

![FIGURE 3 Predicted and actual recruitment. Number of centres open at each time point is also shown.](image-url)
### Table 2

Number of patients recruited by month and centre

<table>
<thead>
<tr>
<th>Month</th>
<th>Oxford</th>
<th>Southampton</th>
<th>Newcastle</th>
<th>Barts</th>
<th>RD&amp;E</th>
<th>Sheffield</th>
<th>Alder Hey</th>
<th>Bristol</th>
<th>RNOH</th>
<th>GOSH</th>
<th>East Lancashire</th>
<th>Nottingham</th>
<th>Stoke-on-Trent</th>
<th>Durham</th>
<th>Leeds</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>October 2014</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>November 2014</td>
<td>0</td>
<td>2</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>2</td>
</tr>
<tr>
<td>December 2014</td>
<td>1</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>2</td>
</tr>
<tr>
<td>January 2015</td>
<td>0</td>
<td>2</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>3</td>
</tr>
<tr>
<td>February 2015</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>March 2015</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>2</td>
</tr>
<tr>
<td>April 2015</td>
<td>0</td>
<td>2</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>2</td>
</tr>
<tr>
<td>May 2015</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>2</td>
</tr>
<tr>
<td>June 2015</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>July 2015</td>
<td>0</td>
<td>3</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>2</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>5</td>
</tr>
<tr>
<td>August 2015</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>2</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>3</td>
</tr>
<tr>
<td>September 2015</td>
<td>0</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>October 2015</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>November 2015</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>3</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>3</td>
</tr>
<tr>
<td>December 2015</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>2</td>
</tr>
<tr>
<td>January 2016</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>2</td>
</tr>
<tr>
<td>Total</td>
<td>1</td>
<td>15</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>6</td>
<td>0</td>
<td>2</td>
<td>2</td>
<td>0</td>
<td>3</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>30</td>
</tr>
</tbody>
</table>

GOSH, Great Ormond Street Hospital; RD&E, Royal Devon and Exeter Hospital.
• At those sites where the PI/surgeons were working well with the team and were fully engaged, recruitment had been most successful and the team were happy.
• Several participants said that showing the study information film\textsuperscript{16} to families was a problem because of a lack of facilities and/or time (not only the study team’s time, but also the families’, e.g. because their car parking time was going to expire).

In July 2015, to get a better idea of the overall DDH patient populations, the SCTU study team asked sites to start completing pre-screening logs. Some sites completed these more successfully than others; however, the logs did report that many DDH cases were successfully treated in a Pavlik harness. In August, the arrangements for an investigator training meeting were abandoned as a result of a lack of response/interest from site teams. In addition, it was decided that Q-QAT involvement would not be used as this involved analysis of actual introductions of the study to families at sites; considering the fact that most sites simply were not seeing suitable patients, this was deemed an unsuitable approach. Therefore, the TMG decided in September 2015 that the trial manager (as sponsor delegate) and the co-CI should conduct individual site visits. A variable response was received when arranging the site visits; visits were successfully conducted at the sites listed in Table 3. The centres that are not listed did not respond.

During the visits, discussion took place with the PI and other members of the team, if available. Each discussion was conducted on a semistructured basis and took approximately 60–90 minutes. All sites were asked to discuss the following topics: (1) current practice (e.g. patient pathway, patient identification strategy, team dynamics, local study management); (2) site-specific issues or practices that might impact the study; (3) key issues inhibiting recruitment at the site, in the opinion of the site team; (4) use of the study information film\textsuperscript{16} and ‘best practice’ of introducing the study to families (as used successfully at Southampton); and (5) study-related training needs (in the site’s opinion).

Below is a summary of the key findings (please note that individual sites have not been identified when a finding is specific to just one or two sites).

**Current practice**
Most sites seemed to work as a ‘joined-up’ team, had good oversight of all clinics and had enough nurse cover.

**TABLE 3** Centres in which visits were successfully conducted

<table>
<thead>
<tr>
<th>Site</th>
<th>Status</th>
</tr>
</thead>
<tbody>
<tr>
<td>Southampton</td>
<td>Not visited as CI’s site</td>
</tr>
<tr>
<td>GOSH</td>
<td>Not visited as co-CI’s site</td>
</tr>
<tr>
<td>Newcastle</td>
<td>Site visit conducted 16 October 2015</td>
</tr>
<tr>
<td>Barts</td>
<td>Site visit conducted 17 November 2015</td>
</tr>
<tr>
<td>Oxford</td>
<td>Site visit conducted 20 November 2015</td>
</tr>
<tr>
<td>Sheffield</td>
<td>Site visit conducted 24 November 2015</td>
</tr>
<tr>
<td>Nottingham</td>
<td>Site visit conducted 24 November 2015</td>
</tr>
<tr>
<td>Bristol</td>
<td>Site visit conducted 6 January 2016</td>
</tr>
<tr>
<td>RNOH Stanmore</td>
<td>Site visit conducted 8 January 2016</td>
</tr>
</tbody>
</table>

GOSH, Great Ormond Street Hospital.
Site-specific issues or practices

One site described a very detailed patient database that was managed by the PI. The PI and team had excellent oversight of all potential patients, which was reflected in the screening and recruitment data from this site.

In contrast, highlighted below are the reasons why some other sites thought that their screening/recruitment may have been lower than expected:

- Screening logs possibly not accurate because of logistic difficulties in recording screening entries as a result of a lack of nurse cover in all peripheral clinics (however, it was highlighted that this would have had little effect on possible recruitment because all surgeons were aware of the study).
- Unless there was a family history of DDH, all babies seen at the clinic were scanned at 6 weeks as a result of a high rate of false positives when scanning at a younger age.
- The 12-week age cut-off age for failed harness is too tight because some clinicians leave the harness on longer and, thus, the harness treatment fails after this age.
- One centre reported that the number of ultrasound hip clinics doubled in 2015; therefore, there is the possibility that DDH detection and successful harness treatments increased, thus possibly treating patients successfully before they become eligible for the Hip ‘Op trial.
- One site stated that for the initial feasibility the entire department was considered, whereas one consultant withdrew their support after this.
- One site said that their standard practice was to wait until 5–6 months of age to operate. As such, it delayed their eligibility assessment until this time, which means that most children had an ON by this age.

Key issues inhibiting recruitment (in the opinion of the local site team)

- Most sites stated that they were keen to do the study but were unable to find enough eligible patients because most were successfully treated in a harness or had an existing ON.
- Some sites suggested that variation in practice could have resulted in few eligible children:
  - Clinician judgement about exactly what constitutes ‘ON present’ – some sites said that they might have been able to recruit more if the protocol had stipulated exactly what constituted ON present on a radiograph instead of allowing sites to choose between this and the more sensitive technique, ultrasound.
  - Some sites suggested that more harness treatments might be successful if the harness is left on for a longer period of time (i.e. some clinicians may choose to treat in a harness for a longer period of time before deciding to operate, thereby reducing the number of potentially eligible children for the study).
  - Some sites faced the challenge of this question from families – ‘what is your normal practice here?’. Some sites felt that when informing families and answering queries about treatment, they felt they could not state certain things because they were not the team’s own experience (e.g. one team felt that they could not tell families that further surgery was more likely if the primary surgery is performed early because this had not been their own team’s experience).

Use of study information film and ‘best practice’ advice

Many sites stated they did use what they believed to be ‘best practice’. Use of the study information video was variable and some sites stated that they did not have facilities to show the information video in the clinic. However, many sites said that they provided the families with the web address for the video so that it could be viewed outside the clinic.

Study-related training needs

Sites did not feel that training was needed or that they could do anything better.
RESULTS

A detailed summary of the observations, actions and outcomes in relation to the management of the recruitment issues encountered during the Hip ‘Op trial throughout 2015 can be found in Appendix 3.

Decision to close the trial early

For most projects with internal pilot phases, the HTA programme holds a face-to-face meeting with the investigators to help it reach a decision on whether or not to proceed to the main trial, the main exception being if an internal pilot has obviously met its progression criteria. This meeting was held for the Hip ‘Op trial on 28 January 2016. The trial team reported that the trial, as specified in the original proposal, could not be delivered in the UK for the reasons outlined previously, but highlighted the exceptional qualitative data and requested that this aspect be explored further. During this meeting, the HTA programme requested that further recruitment to the study be ceased permanently as they agreed that the trial’s primary end point could not be met, but recognised the importance of the qualitative work and invited the trial team to submit a reasonable proposal to allow maximal qualitative data collection and exploitation from the current study.

On 29 February 2016, the study team provided the HTA programme with the following proposal: an 8-month qualitative-only extension that would have allowed completion of the 6-month post-surgery telephone interviews and data analysis for all families already recruited. On 11 April 2016, after careful consideration, the HTA programme informed the trial team that they would like the study to proceed to full closedown without any extension. The programme felt that the amount of additional data was too few to justify the cost of the extension, in addition to concerns regarding participation rates, as the interviews not only depend on the surgery taking place but also on the families agreeing to be interviewed. The funder encouraged the team to pursue alternative funding to complete the qualitative work.

Recruited patients

*Figure* 2 shows the CONSORT diagram for the study. A total of 30 patients were randomised, although one was immediately withdrawn (see Patient withdrawals). Of the 29 patients correctly randomised, 14 were allocated to early treatment and 15 to intentionally delayed treatment. Two patients were lost to follow-up, one from each treatment arm (see Appendix 4). The patient lost to follow-up in the early-treatment arm still provided data up to the 9-month visit, the patient lost to follow-up in the intentionally delayed treatment arm was not seen in clinic after the time of randomisation – the local site team made numerous unsuccessful attempts to contact the family – as such, it has been impossible to ascertain when, or if, the child underwent surgery.

Patient withdrawals

Two of the 30 randomised patients were withdrawn from the study (see Appendix 4). One family, after having agreed to their child taking part in the study, sought a second opinion, whereupon the surgery was conducted in a different centre and not in accordance with the allocated (intentionally delayed) timing. The surgeon who provided the second opinion and performed the surgery was an investigator at a Hip ‘Op trial centre; this incident was fully investigated by the study sponsor. A second child was the subject of a serious breach: the surgeon involved decided to randomise the child on the same day that the child was scheduled for surgery, thus ‘gambling’ that the desired (early) treatment would be allocated. The desired allocation was not assigned at randomisation, the delayed treatment arm was allocated instead, and the surgeon involved was obliged to inform the trial team. This incident was fully investigated by the sponsor and the child was immediately withdrawn from the study.
Patient follow-up

Of the 14 patients randomised to early treatment, all underwent surgery during the running of the trial and were followed up to at least 3 months post surgery by study closure. Of these, five patients also had 6-month follow-up data, five had 9-month follow-up data and one had follow-up data at 1 year post surgery.

Of the 15 patients randomised to the intentionally delayed treatment, eight were known to have undergone surgery by the trial end. Of these eight, two had data at the time of surgery but not beyond that time, one had 6-week follow-up data, one had 3-month follow-up data, one had 6-month follow-up data, two had 9-month follow-up data and one had follow-up data at 1 year.

Numbers analysed

Data are presented based on the 29 patients correctly randomised into the study. All data were analysed using Stata version 14 (StataCorp LP, College Station, TX, USA).

Baseline data and demographics

The majority of patients were aged ≥ 10 months when they were recruited to the study and two-thirds had been treated with a splint before presenting for surgery. As these factors were used as stratification factors in the randomisation, it can be seen that they were well balanced between the treatment arms (Table 4). More girls than boys were recruited to the study, reflecting the prevalence of DDH. The girls were allocated to the treatment arms reasonably evenly, although the boys were not in this very small sample.

Table 5 shows the details of the DDH diagnosis. In the majority of children, it was the left hip that was affected, which is an observation recognised in the clinical literature. The most commonly used (> 80%) imaging technique to diagnose DDH was ultrasound. The median age at diagnosis was 3 months, with the wide interquartile range reflecting patients coming from the failed harness route and the late diagnosis route. The interquartile range for time from diagnosis to randomisation is wide because this also reflects patients coming from the failed harness route and the late diagnosis route.
Surgical procedures

In the early-treatment group, the majority of patients had closed reductions (10 of the 14 including two bilateral procedures), whereas the rest had open reductions. Surgery was performed at a mean 52 days after randomisation [standard deviation (SD) 32 days] and children had a mean age of 210 days (SD 77 days) at the time of the procedure(s). A variety of other procedures were also performed, including adductor tenotomy for 10 patients, gallows traction pre surgery for eight patients, psoas tenotomy for four patients, capsulorrhaphy and acetabuloplasty for one patient, and capsulotomy plus removal of the ligamentum teres for one patient. All patients were treated with a spica cast post surgery.

Of the eight patients in the intentionally delayed treatment group who were known to have had their surgery by trial closure, five had closed reductions and three had open reductions. Surgery was performed at a mean 372 days after randomisation (SD 70 days), and these children had a mean age of 372 days (SD 70 days). Four had adductor tenotomy, three had gallows traction pre surgery, one had psoas tenotomy, two had capsulorrhaphy, three had acetabuloplasty and one had adductor tenotomy; seven of these patients were treated with a spica cast post surgery and the other patient had a frog cast.

Primary outcome

No primary outcome data (presence of AVN at 5 years of age) were collected by the trial closure.

Secondary outcome

Some secondary outcome data were collected by the trial closure, including the presence of the ON at the time of the primary treatment for dysplasia (Table 6) and some information on surgery outcome.

Of the 14 patients randomised to early treatment, surgery outcome data were available only for the 3-month follow-up for three patients, for the 6-month follow-up for five patients, for the 9-month follow-up for five patients and 1-year follow-up for one patient and as such are very limited. At their final reported follow-up,
however, 10 patients reported successfully reduced index hip(s); three reported initially reduced hips at the
time of surgery that then became dysplastic (all three at the 9-month follow-up visit); and one patient
reported a dislocated/displaced hip at the time of surgery that became dysplastic by the 3-month visit.

Of the eight patients randomised to intentionally delayed treatment and who had undergone their surgery
by the trial closure, two had data at the time of surgery but not beyond that time, one had 6-week
follow-up data, one had 3-month follow-up data, one had 6-month follow-up data, two had 9-month
follow-up data and one had follow-up data at 1 year. For the six with post-surgery follow-up, all reported
a successfully reduced index hip.

No conclusions can be drawn from these minimal findings.

**Adverse events**

Six AEs, one of which was a SAE, were reported in five patients. All AEs are listed in Table 7.

### TABLE 6 Ossific nucleus presence at time of surgery

<table>
<thead>
<tr>
<th>ON presence</th>
<th>Time of surgery, n (%)</th>
<th>Intentionally delayed treatment* (N = 8)</th>
</tr>
</thead>
<tbody>
<tr>
<td>ON present at time of surgery</td>
<td>5 (36)</td>
<td>5 (62.5)</td>
</tr>
<tr>
<td>ON not present at time of surgery</td>
<td>8 (57)</td>
<td>2 (25)</td>
</tr>
<tr>
<td>ON present on one side and absent on the other (bilateral DDH)</td>
<td>1 (7)</td>
<td>0</td>
</tr>
<tr>
<td>Unknown ON status</td>
<td>0</td>
<td>1 (12.5)</td>
</tr>
</tbody>
</table>

* Who had undergone surgery during the trial period.

### TABLE 7 Summary of AEs and SAEs

<table>
<thead>
<tr>
<th>Patient ID</th>
<th>Treatment arm</th>
<th>Event description</th>
<th>Start date</th>
<th>Serious</th>
<th>Related to treatment</th>
<th>Severity</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>051008 DT</td>
<td>Artificial</td>
<td>Chicken pox, day 3 of traction prior to closed reduction</td>
<td>24 April 2015</td>
<td>No</td>
<td>No</td>
<td>Mild</td>
<td>Recovering/resolving</td>
</tr>
<tr>
<td>109023 ET</td>
<td>Early</td>
<td>Postoperative pain</td>
<td>8 October 2015</td>
<td>No</td>
<td>Yes</td>
<td>Mild</td>
<td>Resolved 9 October 2015</td>
</tr>
<tr>
<td>109023 ET</td>
<td>Early</td>
<td>Chicken pox, widespread rash, bleeding under plaster</td>
<td>8 November 2015</td>
<td>No</td>
<td>No</td>
<td>Moderate</td>
<td>Resolved 15 November 2015</td>
</tr>
<tr>
<td>549025 ET</td>
<td>Early</td>
<td>Infected birth mark on bottom cause by hip spica</td>
<td>18 February 2016</td>
<td>No</td>
<td>No</td>
<td>Mild</td>
<td>Resolved 25 February 2016</td>
</tr>
<tr>
<td>552015 DT</td>
<td>Artificial</td>
<td>Suspected aspiration during anaesthesia (for planned change of spica cast)</td>
<td>5 November 2015</td>
<td>Yes</td>
<td>No</td>
<td>Mild</td>
<td>Resolved 7 November 2015</td>
</tr>
</tbody>
</table>

DT, delayed treatment; ET, early treatment; ID, identification.
Qualitative aspects

Overview of pilot sample
In-depth telephone interviews were conducted (by SW, a female, experienced, postdoctoral social scientist) with 14 parents (12 mothers and two fathers), 12 of whom were considered the primary carer of the infant who underwent treatment. The key reasons preventing the completion of a larger number of parent interviews are outlined in Table 8 and, primarily, include the non-return of, or a delay in the completion of, demographic questionnaires from the sites.

Families attended clinics across four sites in London (n = 1) and in the south (n = 9), north-east (n = 1) and north-west of England (n = 3). Representing 70% of the pilot target, this recruitment rate is in line with the figures predicted by the qualitative team in the time permitted.

The children of nine of the interview participants had been assigned to the early-treatment arm of the trial, whereas the remaining five had been allocated the ‘intentionally delayed’ arm. Again, the rate of return of the demographic questionnaires determined the proportion of interviews possible with those allocated to the intentionally delayed arm. Four of the 14 patients had an open reduction and the remaining 10 underwent a closed procedure. The average age at which the interview participant’s children had surgery was 7 months (with a range of 4.5 to 13 months) for those in the early-treatment arm and 12 months for those on the intentionally delayed arm (with a range of 11 to 14 months).

Ten of the families had daughters undergoing surgery for DDH and the remaining four had sons. This reflects the increased risk of DDH in girls. Twelve participants lived in dual-parent households and five of the 14 families had other children in 2015. The proportion of participants from minority ethnic backgrounds was thought to be under-represented, although comprehensive data were not collected. The demographic questionnaires asked about the country of birth of the patient and their parent(s), which is not synonymous with ethnic background. In some instances, further information was reported during the interview.

Main findings
The demographic details about participants in the qualitative study are shown in Table 9.

The duration of interviews ranged from 22 minutes and 16 seconds to 98 minutes and 7 seconds, with a mean of 54 minutes. The data were coded (by SW) and co-analysed by Susie Weller and Lisa Roberts. The themes were not specified a priori.

The results outlined below focus on three key areas:

1. parents’ access to, and experiences of, primary and secondary care
2. the impact of surgical intervention for infant DDH on family life
3. participant feedback on involvement in the trial.

### TABLE 8 Key reasons preventing the completion of parent interviews

<table>
<thead>
<tr>
<th>Reason</th>
<th>Count</th>
</tr>
</thead>
<tbody>
<tr>
<td>Interviews completed</td>
<td>14</td>
</tr>
<tr>
<td>Trial patients lost to follow-up/withdrawn</td>
<td>4</td>
</tr>
<tr>
<td>Non-response to interview request before trial closure</td>
<td>2</td>
</tr>
<tr>
<td>Demographic questionnaires not returned from sites</td>
<td>3</td>
</tr>
<tr>
<td>Surgery not completed before trial closure</td>
<td>7</td>
</tr>
<tr>
<td><strong>Total</strong></td>
<td><strong>30</strong></td>
</tr>
</tbody>
</table>
This section focuses on the challenges some families encountered in accessing expert orthopaedic care and/or aftercare advice and support.

Experiences in primary care
Our findings highlight the difficulties some parents faced in gaining a prompt referral from primary to secondary care. Half of the pilot sample \((n = 7)\) encountered either an immediate, and in some instances ongoing, dismissal of their concerns by their GP \((n = 4)\) or some form of delay with the onward referral \((n = 3)\). Of the seven families who described the process as efficient, issues with the health of their infant’s hips were detected for five of them in the labour/postnatal ward:

... pretty much the day after she was born the paediatrician came round the hospital and checked for clicky hips like they do and found a click in her left hip.

Participant 1, mother of female child in early group

... he was born like before midnight, and then he, first thing in the morning they told me that he’s got a hip condition.

Participant 13, mother of male child in intentionally delayed group

### Access to and experiences of primary and secondary care

This section focuses on the challenges some families encountered in accessing expert orthopaedic care and/or aftercare advice and support.

### Experiences in primary care

Our findings highlight the difficulties some parents faced in gaining a prompt referral from primary to secondary care. Half of the pilot sample \((n = 7)\) encountered either an immediate, and in some instances ongoing, dismissal of their concerns by their GP \((n = 4)\) or some form of delay with the onward referral \((n = 3)\). Of the seven families who described the process as efficient, issues with the health of their infant’s hips were detected for five of them in the labour/postnatal ward:

... pretty much the day after she was born the paediatrician came round the hospital and checked for clicky hips like they do and found a click in her left hip.

Participant 1, mother of female child in early group

... he was born like before midnight, and then he, first thing in the morning they told me that he’s got a hip condition.

Participant 13, mother of male child in intentionally delayed group

### TABLE 9 Completed interviews: sample characteristics \((n = 14)\)

<table>
<thead>
<tr>
<th>Child</th>
<th>Interviewee</th>
<th>Duration of interview</th>
<th>Treatment arm</th>
<th>Patient gender</th>
<th>DDH family history</th>
<th>Siblings</th>
<th>Education</th>
<th>Employment</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Mother</td>
<td>37 minutes 27 seconds</td>
<td>ET</td>
<td>F</td>
<td>Y</td>
<td>N</td>
<td>Diploma</td>
<td>Paid FT</td>
</tr>
<tr>
<td>2</td>
<td>Mother</td>
<td>22 minutes 16 seconds</td>
<td>DT</td>
<td>F</td>
<td>N</td>
<td>N</td>
<td>First degree</td>
<td>Paid FT</td>
</tr>
<tr>
<td>3</td>
<td>Mother</td>
<td>48 minutes 47 seconds</td>
<td>ET</td>
<td>F</td>
<td>Y</td>
<td>N</td>
<td>A level</td>
<td>Paid PT</td>
</tr>
<tr>
<td>4</td>
<td>Mother</td>
<td>77 minutes 2 seconds</td>
<td>ET</td>
<td>M</td>
<td>N</td>
<td>N</td>
<td>Higher degree</td>
<td>At home FT</td>
</tr>
<tr>
<td>5</td>
<td>Mother</td>
<td>75 minutes 33 seconds</td>
<td>DT</td>
<td>M</td>
<td>Y</td>
<td>Y</td>
<td>Diploma</td>
<td>Paid FT</td>
</tr>
<tr>
<td>6</td>
<td>Mother</td>
<td>37 minutes 1 second</td>
<td>DT</td>
<td>M</td>
<td>N</td>
<td>Y</td>
<td>Diploma</td>
<td>Paid FT</td>
</tr>
<tr>
<td>7</td>
<td>Mother</td>
<td>40 minutes</td>
<td>ET</td>
<td>F</td>
<td>N</td>
<td>N</td>
<td>First degree</td>
<td>Paid FT</td>
</tr>
<tr>
<td>8</td>
<td>Mother</td>
<td>98 minutes 7 seconds</td>
<td>ET</td>
<td>F</td>
<td>Y</td>
<td>Y</td>
<td>First degree</td>
<td>Paid FT</td>
</tr>
<tr>
<td>9</td>
<td>Mother</td>
<td>49 minutes 58 seconds</td>
<td>ET</td>
<td>F</td>
<td>Y</td>
<td>N</td>
<td>A level</td>
<td>At home FT</td>
</tr>
<tr>
<td>10</td>
<td>Mother</td>
<td>33 minutes 48 seconds</td>
<td>ET</td>
<td>F</td>
<td>N</td>
<td>Y</td>
<td>Vocational</td>
<td>Paid FT</td>
</tr>
<tr>
<td>11</td>
<td>Father</td>
<td>44 minutes</td>
<td>ET</td>
<td>F</td>
<td>N</td>
<td>Y</td>
<td>AS level</td>
<td>Paid FT</td>
</tr>
<tr>
<td>12</td>
<td>Mother</td>
<td>81 minutes</td>
<td>DT</td>
<td>F</td>
<td>N</td>
<td>N</td>
<td>First degree</td>
<td>Paid PT</td>
</tr>
<tr>
<td>13</td>
<td>Mother</td>
<td>50 minutes</td>
<td>DT</td>
<td>M</td>
<td>N</td>
<td>N</td>
<td>A level</td>
<td>At home FT</td>
</tr>
<tr>
<td>14</td>
<td>Father</td>
<td>55 minutes</td>
<td>ET</td>
<td>F</td>
<td>N</td>
<td>N</td>
<td>Professional</td>
<td>Paid FT</td>
</tr>
</tbody>
</table>

A level, Advanced level; AS level, Advanced Subsidiary level; DT, delayed treatment; ET, early treatment; F, female; FT, full-time; M, male; N, no; PT, part-time; Y, yes.
Two of these seven parents experienced a prompt referral through other means. Concerned about her daughter, participant 7 was referred for further tests after a thorough examination by a locum GP, who requested an urgent X-ray because of the child’s age. Alternatively, participant 14’s daughter was advised to seek tests by a chiropractor who was treating his daughter for another condition.

Four parents described a catalogue of issues with gaining a referral that primarily centred on the dismissal of their concerns by their GP:

It was really disheartening because I knew . . . I just felt like I was going mad because I knew there was something wrong but they [doctors] were telling me that it was obviously fine.

Participant 5, mother of male child in intentionally delayed group

Similarly, participant 2 visited her GP on five occasions expressing concerns about her child’s leg length, hoping for an onward referral:

My problem was I wasn’t going in there saying there’s something wrong. I don’t know what. We were pointing out that we thought the problem was her legs . . . the difference in her leg length. The creases were at a different stage on her leg.

Participant 2, mother of female child in intentionally delayed group

She, along with two other parents whose concerns had been disregarded in primary care, had a family history of DDH, which was also not acknowledged as a risk factor.

I took her to my GP at 6 weeks. Um . . . again I had to explain the history and again she was like ‘Oh I can’t see or hear anything. It all sounds fine’. And we really had to push for a scan for our daughter . . . we just really . . . we kept just explaining to people how strong the family history was but we didn’t seem to really be um . . . I suppose people weren’t really taking us seriously.

Participant 3, mother of female child in early group

On the recommendation of a child care professional, participant 5 eventually obtained a referral from her local health visitor, although she also encountered some issues accessing this weekly service as she worked on a full-time basis.

The pilot sample also contained examples of antenatal concerns, based on a family history of DDH, that were dismissed by midwives and maternity care practitioners resulting in late diagnosis and significant distress, as the following examples highlight:

I think in my maternity notes the midwives, doctors sort of anyone that would listen really I did explain the . . . the strong family history. My daughter was born at the hospital . . . and I’d spoken to two sort of paediatric doctors that would check her over and they just said ‘oh the hips aren’t making a clicking sound. It sounds OK’.

Participant 3, mother of female child in early group

I kept saying, throughout pregnancy, to my midwife that something wasn’t right . . . they didn’t follow through. They didn’t follow-up on what I was concerned about because I kept saying there was sort of a lump that didn’t feel quite right. I went into labour and the midwife thought he was breech.

Participant 6, mother of male child in intentionally delayed group
For three parents, even an early first assessment did not necessarily result in a prompt follow-up or a straightforward path to treatment. Issues with participant 8’s daughter’s hips were detected at birth, but with delays to the processing of appointments she had to push for a new referral:

> We never got anything through um, about her appointment, so I spoke to the health visitor and to the midwife, um, none of which seemed to know how we were supposed to follow that up. In fact, I was told to leave it, I think . . . you know, ‘cause 6 to 8 weeks, and then if we hadn’t heard anything then we should have gone through to our GP, and then got re-referred.

Participant 8, mother of female child in early group

Similarly, participant 12’s original referral was overlooked:

> . . . when I um, saw my health visitor, I raised it with her that the doctor had said that she had a clicky hip, um, and then they started chasing the appointment. It turns out the um, doctor hadn’t actually referred my daughter for a scan.

Participant 12, mother of female child in intentionally delayed group

The repeated disregard for parents’ concerns about their child’s hip health/physical development caused much anguish for families, as summarised by participant 2:

> . . . it’s been an emotional rollercoaster to be honest because we took her to be GP quite a lot when she was younger pointing out that we thought there was something wrong with her legs and they said that there wasn’t and so then to go to the diagnosis . . . diagnosis of hip dysplasia was quite a shock actually.

Participant 2, mother of female child in intentionally delayed group

**Surgery experiences**

Despite the difficulties over half the sample faced in accessing expert orthopaedic care, parents generally spoke positively about their experiences of their child’s surgery, with 13 suggesting that they were either ‘likely’ or ‘extremely’ likely to recommend the orthopaedic department in which their child was treated to friends and family, with one less decisive. All participants felt they had had time to discuss the surgical options and surgeons were held in high regard for the expertise and skills in clinical procedures, as the following examples from different sites illustrate:

> I think we’re amazed with what they [the surgical team] can do . . . and very yeah, very, you know, speak very highly of what they do.

Participant 4, mother of male child in early group

> They’ve [surgical team] all been absolutely fantastic. Our surgeon is a brilliant surgeon.

Participant 6, mother of male child in intentionally delayed group

> I’m very pleased with my experience . . . it was a troubling time for us but they [surgical team] put us at ease.

Participant 7, mother of female child in early group

Parents felt informed about the likely clinical procedures they would encounter, although this was not necessarily the case for other aspects of the process (see, for example, Aftercare advice and support):

> The surgeon, you know, he was, he was crystal clear on what was going to ‘appen and stuff, so you know, we were under no illusions as to what was going to happen; we just didn’t have any information on how we’d cope with it.

Participant 11, father of female child in early group
Several parents suggested that the limited availability/accessibility of those with clinical expertise to answer any queries was one area that could improve patient experience, as elucidated by participant 12:

*You probably just could have done with maybe somebody back up on the ward – even one of these nurses back up on the um, day ward, just to talk through a few things with or even the surgeon coming to see you before he left for the day.*

Participant 12, mother of female child in intentionally delayed group

Despite the esteem with which surgeons were held, other aspects of the process, particularly experiences of referral and aftercare (see *Experiences in primary care* and *Aftercare advice and support*), were not described in such positive terms:

*I think the only thing I would say is that we . . . on the few different visits we got very different stories, and it was very hard to work out what was going on.*

Participant 8, mother of female child in early group

When asked about their expectations of the surgery before the operation, some spoke about uncertainty and the fear that the intervention would not be successful, as the following extracts demonstrate:

*. . . where I had a closed and it didn’t work and I just thought . . . I don’t know . . . I’d read a lot as well because there’s a Facebook [Facebook, Inc., Menlo Park, CA, USA] site for hip dysplasia and a lot of people on there have had a similar experience that their child’s had to have one lot of surgery and then another lot of surgery and I kept thinking ‘oh god what if that happens to us’.*

Participant 11, mother of female child in early group

*. . . I wasn’t expecting it to be very nice . . . I think it’s quite scary well it was for us anyway.*

Participant 2, mother of female child in intentionally delayed group

*I didn’t really have a clue what to expect because we were told that his case was a very severe case of hip dysplasia . . . we weren’t sure whether it was going to work . . . or whether it could work temporarily and then it could possible move again so we weren’t entirely sure . . . sort of went in with our eyes shut.*

Participant 6, mother of male child in intentionally delayed group

Among the expectations were very pragmatic hopes that one surgical intervention would rectify the problem:

*She might need further surgery along the line but really I’d like it to fix her.*

Participant 3, mother of female child in early group

Parents explicitly spoke of the shock they experienced seeing their child or other children either in traction or in a cast for the first time:

*. . . you just always don’t expect your child to come out in a cast like they’re going to . . .*  

Participant 2, mother of female child in intentionally delayed group

*After the surgery it was the shock of seeing him in this cast . . . to sort of see him like that . . .*  

Participant 4, mother of male child in early group

*. . . the cast’s an absolute monstrosity. It’s just . . . it’s just a massive thing hanging off ‘er.*  

Participant 11, father of female child in early group

*We met another family . . . who were essentially a week ahead of us. So we could see their daughter in the traction, which um . . . being completely blunt is a little bit of a shock/worry for your child.*  

Participant 14, father of female child in early group
Post surgery, many parents expressed a sense of relief that the intervention had not been as bad as they feared and that their child required less invasive surgery than they had originally imagined:

*It all went well to be honest. They were very good even putting her to sleep to coming round they were very good. They were very good and how to care for the cast... it was... it was good.*

Participant 2, mother of female child in intentionally delayed group

*... the fact that they didn’t need to do an open... the surgeon was quite hopeful that he wouldn’t have to and he didn’t so the fact that the hip went back in without the need to open stuff up.*

Participant 3, mother of female child in early group

*I actually expected them to have to go... they didn’t actually have to cut in or anything. I expected it to be a lot more severe than it was... so when she came out um... I was pleasantly surprised.*

Participant 7, mother of female child in early group

*. . . well to be honest the whole thing was all worth it in the end. The stay in there for 2 weeks in, while she was in traction . . . but it all, in the end, I would ... well so far it’s been successful, and it’s all worked out really well.*

Participant 9, mother of female child in early group

Those who had the opportunity to talk to other families further along the process were much more able to manage their expectations.

For many, plaster technicians provided valuable advice on aftercare that was somewhat lacking elsewhere (see Aftercare advice and support). Nonetheless, a small number of parents felt traumatised and confused by the advice given about the drying of the cast immediately after application. One mother described, in detail, how the guidance provided by hospital staff contradicted more general information about the safe care/positioning of infants:

*. . . she had to go on her front because the back of the cast needed drying... but because she was only just 6 months she wasn’t used to being on her front... and I explained that to them but she was on her front on sort of loads of pillows... and because she was getting so distressed from being on her front she kept shoving her face in the pillows and you could hear her not... couldn’t breathe properly. Yeah and they shouldn’t be left. I said to the nurse, you know, how long does she have to be like this for and she said ‘well at least for a couple of hours because the cast needs to dry’, but I said but I said ‘she can’t even hold her... she can just about hold her head up. She’s not used to being on her front. She stuffing her face in the pillows. She can’t breathe’.*

Participant 1, mother of female child in early group

This instruction proved distressing for both the parent and child. A similar encounter was experienced by participant 12:

*. . . the day after the op[eration], or sort of the evening, like, she’s had her op, she was in cast and we had to sort of flip her back and forth to help the cast dry... um, and the nurses said that they would do that for me overnight... they didn’t, I think they missed it.*

Participant 12, mother of female child in intentionally delayed group

Although structural issues with the fabric of the cast were rare, two parents described having to make multiple return visits:

*. . . when he came out of surgery at that point he needed sort of sleek [tape] to be put on and um... padding and stuff to take away sharp edges um... and for it to dry out properly. Um... which these people didn’t know how to do... or what it was that we were wanting... because we weren’t able... to understand what to do.*

Participant 12, mother of female child in intentionally delayed group
to sort of stay long enough for the cast to dry the second time round then it collapsed slightly around the bum area. We had to go back the next day, which was a nightmare.

Participant 4, mother of male child in early group

... the first one it kept sort of softening ... and it looked like it was breaking so we were back and forth to A&E [accident and emergency] at night after work um ... which was just really tiring and then having to be awake with her in the night and then back to work again in the day.

Participant 7, mother of female child in early group

For participants 4 and 7, return visits included difficult or lengthy journeys.

Aftercare advice and support

During the interviews, parents were asked about the activities of daily living and of the particularities of looking after a child for a minimum of 12 weeks in a waist–ankle hip spica plaster cast. In addition to concerns about the fragility of the cast, especially if exposed to moisture, personal hygiene was a significant and common theme. Parents of infants in a hip spica are tasked with daily ablutions but cannot bathe their child easily. Parents spoke of the challenges faced and the techniques they developed, often by trial and error:

... it’s difficult actually. You’ve got to clean her very, very well. It’s not as easy as just changing her nappy ... It’s quite difficult when they’re out actually because she’s big just to put her on a normal changing table is quite ... isn’t great to be honest so yeah it has been difficult.

Participant 2, mother of female child in intentionally delayed group

... when you washed her on a night and then it was a bit tricky trying to push the, like had a poo or anything like that up her back couldn’t get your hand all the way up the plaster, so it was very tricky.

Participant 10, mother of female child in early group

Of concern was the possibility of the deterioration of the cast by excrement, for which parents had to find their own ways of refreshing the padding:

Oh that was tough because she went into her first spica cast and had really bad diarrhoea ... for 6 days so the cast got absolutely ruined ... so I had to take out all of the wadding that was already in there and replace it with sort of fresh cotton wool.

Participant 3, mother of female child in early group

We did have a few leaks but we sort of were able to rectify, um, and that, but that was the other thing: I was ... at the time she was having leaks, and quite early on there was a couple of accidents ... nothing major but I would take out the lining that was in the um, tsk, in the plaster, like the um, it was pretty much like cotton wool stuff.

Participant 12, mother of female child in intentionally delayed group

Although all parents felt the surgical procedures had been thoroughly outlined, a lack of access to aftercare advice and support was a recurring theme across the interviews. This was particularly apparent for hygiene issues, such as nappy changes, made even more challenging by the presence of a hip spica cast:

I got shown very quickly afterwards how to change a nappy and that was it, but it was things like how do I wash her hair, and advice or what I can use. Just practical everyday things ... There didn’t seem to be as much information.

Participant 7, mother of female child in early group
I probably’d have liked to have known more about how, like, she would have, like, dealt with being in the cast afterwards, and ... um, what, what, like, were the best positions to put her in, and stuff like that; they didn’t really ... the hospital didn’t really tell us enough about what was more suitable for her, what wasn’t; it was just sort of like she’s in a cast, and that, you know, just take nappy in and do the nappies and that’s it.

Participant 9, mother of female child in early group

... we felt like we didn’t get enough support in terms of how to hold, or carry, or change her or, ‘cos like you just sort of got to find your own way through it.

Participant 12, mother of female child in intentionally delayed group

Of frustration was a lack of access to expert aftercare support while in hospital, resulting in some using their own limited experience to guide health-care professionals:

... I think what, what struck me as a parent – it’s not a criticism of any of the nurses – but um, like some of the nurses really didn’t seem to know how to deal a nappy situation ... and I think as a parent you’re like, my god! It’s like two nurses are struggling together to do this nappy ... how the hell am I going to be managing it on my own ... then I had this amazing nurse came round, and oh my god! It was like a formula 1 tyre change once she was ... so, like, she had it down pat; this woman was amazing.

Participant 8, mother of female child in early group

... after the surgery, the aftercare we got on, on the ward was appalling; it was d-, it was absolutely dreadful. You know, we asked a nurse to show us how to do our daughter’s nappies, she went ‘oh I don’t know’. And I went ‘well if you don’t know how the hell am I supposed to know?’.

Participant 11, father of female child in early group

... one of the nurses was really incompetent, and um, she came over to change her, um, but she didn’t know, like, I’d sort of read up on it, and maybe had had a conversation with another nurse earlier in the day, um, and had changed her nappy with her, and like you have to cut the tabs off and wedge it up inside the cast ... and this new nurse that was on for the night shift didn’t know that, and she was just gonna try and get this nappy on, I don’t know how, but I said to her, ‘You need to take the tabs off’. So I think I ended up showing her.

Participant 12, mother of female child in intentionally delayed group

I asked them to show me how to change the nappy so I wouldn’t damage the cast, or I wouldn’t hurt him ... and I would say from all that nurses only ... one knew, and I could not get her. So it was like ... a beginner who showed me how to change the nappy.

Participant 13, mother of male child in intentionally delayed group

Shortcomings in aftercare advice were apparent across three of the four sites. One key issue for parents was either contradictory information or discrepancies between the (limited) advice given by professionals and the everyday lived experience of what works in practice at home:

I’d say each nurse had differing opinions of how to care for the cast, so one said to dry her constantly when changing her nappy. Someone said to use talc and to be honest I didn’t have a clue ...

Participant 2, mother of female child in intentionally delayed group

For some, this related to the challenges of reconciling clinical procedures necessary for the healthy development of the hip and the practicalities of caring for a child in a cast. Participant 4, for instance,
recognised the role of the spica cast in her child’s treatment but wished for adaptations to the structure (i.e. a larger hole for the nappy) to aid care:

... we appreciate that they’re there to do the job correctly and for it to sit correctly but I think it’s also just remembering the kind of day-to-day care that we’ve got to do ... whether it’s that size of the hole also um ... um ... after ... I know after the first cast you don’t tend to get the coloured coating on the outside the cast ... which actually we found acts as a lot um ... as a sort of more of a smoother barrier [more comfortable for the carer].

Participant 4, mother of male child in early group

Some compensated for the shortcomings of aftercare support by drawing on the advice given by a key third-sector organisation, Steps, although this was not a common approach among the pilot families.

The impact of surgical intervention for infant developmental dysplasia of the hip on family life
The impact on families of surgical intervention for infant DDH, including the timing of procedures, remains underexplored. This section examines a broader range of effects from the implications for family finances and paid employment, through to parental well-being and the responses of siblings. The diversity of experience will be highlighted.

Finances and resources
The pilot study revealed the high personal costs associated with DDH treatment including extended parental leave, plus annual, emergency and unpaid leave for hospital appointments.

Impact on paid work and parental leave  One key impact of the timing of surgery for infant DDH was on paid employment, particularly parental leave. The interviews explored workplace support for/barriers to parents returning to work from maternity or shared parental leave, along with requests for additional leave for appointments and hospital stays. These facilitators and barriers have significant implications for both the timing of surgery and family-friendly employment policies. The most recent survey of maternity and paternity rights and women returners found that the average length of maternity leave taken in the UK was 39 weeks in 2008, which coincides with the paid period of leave.19

From the pilot interviews, the families’ approaches to incorporating treatment and rehabilitation into their home and work lives may be stratified as those with flexibility and resources, professional families reliant on kin care, mothers exiting the labour market and financially constrained families.

Type 1: flexible resourced families  Four of the 14 families may be described as flexible in their approach to incorporating surgical treatment and rehabilitation into their lives. Although all families exhibited some degree of flexibility, these families had the capacity to make choices and adapt life around the condition. They reported a range of household incomes, but tended to be at the higher end of the spectrum. Before having children, each mother had worked on a full-time basis but had the resources necessary to extend their maternity leave. They, or their partners, were able to engage in flexible paid work enabling them to combine home, work and care tasks.

For example, participant 1 and her partner both worked on a full-time basis. Her daughter had surgery at the age of 6 months (early-treatment arm) and she extended her maternity leave to accommodate the period of treatment and recovery:

I was on maternity leave but I was due to go back ... when did ... she had her operation in January and I was due to go back sort of February time ... but I extended that until the end of March.

Participant 1, mother of female child in early group
When she did return to work, she was able to negotiate a shared pattern of paid work and care with her partner by working in the evenings and at weekends. In the first two parental cost diary entries, at the time of surgery and the 6-week follow-up, she stated that the situation had affected her work in a bad way but by 6 months post surgery the effect on work had subsided. Her partner used annual leave (11 days noted across the diaries) to attend appointments:

...the other half had to take quite a few days off for sort of for hospital and... but mainly used his holiday because otherwise he wouldn’t get paid and we’d be down quite a lot of money.

Participant 1, mother of female child in early group

Similarly, participant 3 worked part-time and her partner was engaged in shift work on a full-time basis, both in skilled professions. She had previously been occupied in a full-time managerial position but had returned to a lower grade post after maternity leave. Her daughter had surgery at the age of 6 months (early-treatment arm) and she elected to extend maternity leave into the unpaid period of entitlement to accommodate rehabilitation:

I was going to take 9 months off ... and go back in sort of May/June time with a bit of holiday attached on ... I chose then to take a year ‘cos you don’t get paid for the last 3 months ... so I’m not being paid at the minute. I chose to take that for where her cast is going to come off ... but also I’m kinda glad that I have because it’s given me a bit more time with her so although I chose it for a different reason I am enjoying the extra time off with her.

Participant 3, mother of female child in early group

In this respect, she did not feel the situation had negatively impact on her work. Her partner had taken some annual and emergency leave, and by working shifts he was able to accommodate appointments into his working day:

He works shift work so quite often appointments will fall when he’s off so for example he’s got a week off now so um there is a bit of flexibility there with his shift patterns.

Participant 3, mother of female child in early group

Participant 3 had also experienced positive communications with her employer about additional time away from work to accommodate follow-up appointments.

A further example is that of participant 8 and her partner who both work full-time in professional positions. Their daughter had surgery at the age of 6 months (early-treatment arm). She had extended her intended period of maternity leave to cover her daughter’s surgery and rehabilitation:

I extended it, so I probably would have gone back a little bit earlier, but I wanted to see her all the way throughout the process ... Well I was lucky because where she was selected to go on the um, the early one [treatment arm] to the 6 months ... I was on maternity leave, so I didn’t go back to work until um, a week after she came out of broomsticks.

Participant 8, mother of female child in early group

Being able to combine the period of treatment and recovery with her maternity leave reduced the potential intrusion of the process on her work life and care arrangements:

I would be there to be able to be that carer for her all the way through without it impacting on nursery or grandparents or anybody else.

Participant 8, mother of female child in early group

She spoke of being concerned about how they might have managed their home–work life should surgery have taken place after her daughter reached the age of 1 year, and had concerns around the willingness...
of formal child care providers to accommodate a child in a cast (see Impact on child care options). They had contemplated the options, including unpaid family leave, should the surgical intervention have been scheduled for after her return to work.

The family had some flexibility in their care arrangements as participant 8’s partner worked a rolling shift pattern, otherwise she was supported by her mother-in-law:

... he works like a rolling shift pattern ... so for appointments he could come to it; some he couldn’t, so when he didn’t attend with me, my mother-in-law came with me instead ...

Participant 8, mother of female child in early group

The child’s father was, however, denied compassionate leave for the time his daughter was in hospital.

The final example of a ‘flexible family’ is participant 12, who worked in an intermediate profession. She did not disclose her partner’s job but noted that he worked full-time. Her daughter had surgery at the age of 11 months (intentionally delayed treatment arm). She elected to extend her maternity leave to cover the time her daughter was in a cast:

[I was due back in] ... middle of December, um, and then the operation was booked for end of November, so at which point they agreed to um, parental leave. So I took con-, er, I was on parental leave, unpaid leave, um, from then to my return to work, which was April.

Participant 12, mother of female child in intentionally delayed group

She therefore used her unpaid maternity entitlement, which, as she was the highest earner in the household, had financial implications for the family. Her partner took annual leave every time their daughter had an appointment, particularly during their hospital stay.

Participant 12 did not believe she had received a positive response to her requests for leave from her employer, suggesting that she had been treated unfairly. She had missed one of her contact keeping in touch days while on maternity leave as a result of the fitting of her daughter’s Pavlik harness, which she felt was later held against her:

I told them I wasn’t going in, and I told them why. Er, then that’s later been used against me, um, not in a big way, but it’s sort of been thrown back at me that, ‘Oh, you had to cancel um it’s as though you had to ... you’ve had to ... you had to say “no” to a lot of work because of your daughter’.

Participant 12, mother of female child in intentionally delayed group

The ‘flexible families’ not only had the resources to extend periods of leave but also had the flexibility within their employment situations to fit work life around their child’s condition and treatment. Although this was not necessarily an easy task, they did not tend to report significant financial implications.

Type 2: professional families kin care The two examples in this category are of families that managed the pressures of balancing work and family life through the period of surgery and rehabilitation with extensive support from their extended kin/families. In both instances, the participants worked full-time in professional positions. Participant 2 and her partner, for example, both work on a full-time basis, in professional and skilled work. Their daughter had surgery at the age of 11 months (intentionally delayed treatment arm). Daily child care was provided by her mother and mother-in-law during rehabilitation as she’d rejected formal child care provision (see Impact on child care options):

... my mum and my mother-in-law, they both look after her so it’s kind of a swap and change but yeah it’s had quite an impact.

Participant 2, mother of female child in intentionally delayed group
In combination with care provided by kin, she relied also on using her (unpaid) parental leave entitlement:

*My partner gets a lot more holidays than what I do, which is why I took the unpaid work but they were very flexible actually in letting me take as much leave but I’m entitled to a week unpaid anyway for parental leave … just because of the child’s so young.*

Participant 2, mother of female child in intentionally delayed group

In her diary entries, participant 2 reported taking a total of 18 days paid/unpaid leave, and her partner took 10 days, some of which was paid annual leave. She stated that the situation had a detrimental effect on her work and the strains of juggling a professional role and concerns about kin care were apparent.

Similarly, participant 14 is a full-time professional. He had separated from his partner, who is self-employed and worked on a part-time basis. Their daughter had surgery at the age of 10 months (early-treatment arm). She lived part-time with each parent and on weekdays was cared for by her mother, aunt and parental grandparents alternately. Indeed, participant 14 relied on care from family members to enable him to engage in paid work. He acknowledged the level of care and the challenges of handling a child in a cast that he was expecting elderly members of the family to undertake:

*There’s all these things that you know but until you’re actually doing it um … and also you’re suddenly asking, OK family member, but you’re asking them to take a higher level of care.*

Participant 14, father of female child in early group

Both parents also had some degree of flexibility in their work life. Participant 14 was in a senior position and able to manage his own time:

*We’re fortunate enough to have a little bit of flexible working. As I say I was sort of managing it … you know, if I had to take some hours I would give some hours back.*

Participant 14, father of female child in early group

They continued to work while also engaging in intensive periods of care for their daughter on the basis that they might need to take leave in the future if elderly relatives were not able to manage to look after her:

*… the week that she was in traction was quite … is it difficult? I suppose we were still trying to manage our own work situations and we were sort of doing 24-hour shifts with our daughter. One on, one off sort of thing.*

Participant 14, father of female child in early group

These two families relied on kin care to enable them to manage the demands of professional and more senior job roles. These two cases highlight the stresses involved in continuing to work through the period of rehabilitation and the challenges of being flexible during hospital stays and for appointments.

**Type 3: labour market exit mothers** The four parents (or, in one instance, their partner) in this category have all left the labour market – either in the short or long term – to care for their children. Unlike the flexible families who had extended their maternity leave, these families no longer had a job to which they could return. When reported, they reside in a wide range of circumstances from those with a partner in a senior professional position to a lone mother. Becoming a full-time carer was not always presented as a choice but a necessity.

For example, participant 4, a former freelance designer, had elected to become a full-time carer for her son after he was diagnosed with DDH. Her partner had a full-time professional job. Her son had surgery at
the age of 7 months (early-treatment arm). She opted not to return to paid employment until her son had fully recuperated and was out of his cast, describing herself as currently ‘unemployed’:

“I’ve been unemployed and now I’m sort of I’ve known the whole [time] while my son has been in his cast that, you know, I couldn’t get a job because my son needed full-time care and um it wasn’t a case . . . we didn’t feel comfortable knowing that if he was to go to nursery or something he wouldn’t have a one-to-one person, which we felt he needed with his condition.

Participant 4, mother of male child in early group

She had not intended not to return to employment and believed the situation had had a negative impact on her career. She also noted implications for her partner managing his job on a day-to-day basis during treatment:

. . . my husband at work sort of concentration-wise it was quite hard for him, you know, and his boss could definitely see afterwards what a weight had been lifted . . . and um . . . how much better he was able to focus.

Participant 4, mother of male child in early group

As her partner had used all his annual leave for DDH-related appointments, the family was unable to carve out any respite time in the form of a holiday. He also had to take unpaid leave, which had some financial implications. She called for greater opportunities to take compassionate leave in such circumstances.

Similarly, participant 9 is a full-time carer for her daughter. Her partner works as a skilled manual worker. Her daughter had surgery at the age of 7 months (early-treatment arm). Like other mothers in this category, she exited the labour market as she did not feel that her child would be cared for appropriately by an external provider:

. . . the only thing that has changed is that I would’ve gone back to work a lot sooner, but um, as she got put in the cast and she was medically, you know, in ways unwell I suppose with her hips, I couldn’t go back as soon.

Participant 9, mother of female child in early group

She felt that she could not return to work until after the period of rehabilitation and could not expect her partner to take any leave as his income was vital to the household finances. On track for promotion, she stated that her partner was worried that any period of leave would jeopardise his chances of an increase in pay, despite his desire to be present at appointments for his daughter:

He wanted to be able to go to work every day, obviously, ‘cause we had rent to pay and things, but then some days I’d have to say, look that date’s coming up so you have to take that off and he’d worry about the bills and oh god! I should be working but I need to be there.

Participant 9, mother of female child in early group

In the future, she planned to get part-time work at the weekends, thereby providing continued flexibility.

Participant 11 was a father who was employed full-time in an intermediate profession. His partner had left her part-time job to care for their daughter who had surgery at the age of 4.5 months (early-treatment arm). As a result, the family had lost part of their household income and she had forfeited her career; a decision made by the couple as her income was lower:

. . . she wasn’t very ‘appy about it because she didn’t want to be a stay-at-home mum . . . you know, she wanted to be a working mum.

Participant 11, father of female child in early group
Participant 11’s role in the public sector meant that he had to apply to senior managers for special permission to take leave during his daughter’s time in hospital. Initially, his request was rejected on the grounds that the reason was not significant enough to have an adverse day-to-day impact on the participant:

\[\ldots\text{initially got rejected, um, ‘cause it got past the\ldots he said it wasn’t an emergency and, and anything significant happening in your life.}\]

Participant 11, father of female child in early group

Leave was eventually granted.

Finally, participant 13 is a full-time carer for her son. She is a lone parent. Her son had surgery at the age of 11 months (intentionally delayed treatment arm). She had decided not to return to work after maternity leave and to, instead, care for her son, opting out of formal child care provision:

\[\ldots I\text{ was planning to come, to go back to work just after my maternity finished, but then I didn’t want to put him into the nursery, even after being told that he can go \ldots but, I just say I couldn’t.}\]

Participant 13, mother of male child in intentionally delayed group

She stated that she had received some advice from her local job centre that suggested that she could be a full-time carer for up to 2 years.

The mothers in this category all exited the labour market, albeit temporarily, to care for their children as they did not consider external child care provision appropriate. The decision for the mother to leave her position was, in part, determined by her salary/contribution to the household. Underlying such ‘decisions’ are also gendered expectations around parenting.

**Type 4: financially constrained families** In the four families in this category, both parents worked full-time and reported a household income of < £30,000 per annum. Three of the children attended formal child care provision on a full-time basis, whereas one was cared for, in part, by her unemployed father.

Participant 5 and her partner both work full-time as skilled and semiskilled manual workers. Her son had surgery at the age of 14 months (intentionally delayed treatment arm). She had to return to work for financial reasons. She expressed feeling conflicted over her annual leave as her vacations needed to coincide with the school holidays of her older children, rather than her youngest son’s treatment. Her partner took a few days’ annual leave, but had just started in a new position and found it difficult to take time off. Her workplace was unable to help with additional paid leave and advised that she seek guidance from her GP, who granted her sick leave for the period:

\[\ldots\text{the reason I had to do sick was because my work don’t pay sick if your child is sick \ldots because obviously my son was ill. It was his \ldots it was his operation and stuff like that \ldots Um \ldots I couldn’t \ldots I wouldn’t have been paid. They said to me ‘We can give you emergency leave. That’s not a problem’}.\]

Participant 5, mother of male child in intentionally delayed group

Participant 5 simply could not afford to take any unpaid leave. The time negotiated with her GP was, however, complicated by two postponements of her son’s surgery (one because of sickness and another because of an administrative error), which resulted in her having to reorganise timings:

\[\ldots\text{and it was \ldots it was very emotionally draining and I’ve got to wait another week. I’ve got to \ldots I’ve got to go back to work. He’s got to go back to work with me}.\]

Participant 5, mother of male child in intentionally delayed group
She reported that her workplace had been very understanding in providing flexible work hours for
appointments, but the stress of the negotiations was apparent.

Participant 6 and her partner both work full-time in skilled non-manual positions. Her son had undergone
surgery at the age of 1 year (intentionally delayed treatment arm). She had recently been promoted, which
provided her with greater opportunities for working from home. She spoke of having to return to work
after 9 months of maternity leave for financial reasons:

\[ I \text{ didn’t go back to work until he was 9 months old. It . . . obviously once I did go back to work it
going back and forth to hospital I was having to take days off. And again the same with my partner.
I wasn’t actually going to go back to work because of it. Um . . . I physically had to due to
financial reasons.}\]

\[ \text{Participant 6, mother of male child in intentionally delayed group} \]

She used annual leave accrued during her maternity leave for appointments and hospital stays, with
9 days’ leave mentioned in the 3-month follow-up diary entry. Her son’s condition had had a negative
impact on her work in the period before surgery, but by her son’s 3-month follow-up visit this
had receded.

Participant 7 and her partner are both full-time skilled non-manual workers who each undertake a
long commute from home to work. Her daughter had undergone surgery at the age of 13 months
(early-treatment arm) and was in full-time paid child care provision. She spoke of the stress and anxiety
she felt requesting time away from her new job, for which she had been granted compassionate leave:

\[ I \text{ hadn’t really returned. It was . . . it was a new job. I was obviously feeling very stressed. I was very
anxious. I had a new job and I had to ask for all this time off. I’m just very fortunate that they’ve been
so good.}\]

\[ \text{Participant 7, mother of female child in early group} \]

Her partner had had to use annual leave for appointments, which left him without any of the vacation
time vital for respite and recuperation. Both had received much support from their employers, enabling
them to take time off:

\[ \text{. . . our work has been very, very good. Very accommodating um . . . we’re very fortunate where we
work that they let us go um . . . whenever we need to . . .}\]

\[ \text{Participant 7, mother of female child in early group} \]

Her anxiety had been fuelled by having to leave work urgently on DDH-related matters, sleep deprivation
while her child was in a cast, and long commutes between home, work and the hospital.

Finally, participant 10 worked full-time in an unskilled manual position. Her partner was unemployed and
at home full-time. Her daughter had surgery at the age of 6 months (early-treatment arm). She attended
the pre-surgery hospital appointments while on maternity leave and used her annual leave entitlement for
other hospital appointments. She had not extended her intended maternity leave. She did not feel that the
process had any impact on her job and had found her employer very supportive:

\[ \text{They were understanding and stuff, and they let us have the time off to go to the hospital for her.}\]

\[ \text{Participant 10, mother of female child in early group} \]

The resources available to families, coupled with the circumstances in which they are situated, are
implicated in the ways in which they manage the process of treatment and rehabilitation.
Impact on child care options

The majority of parents were not comfortable contemplating formal child care, especially institutional settings, when their child was in a cast. In some instances, local provision simply did not cater for the specialist care required:

... we haven’t been able to get child care for her ‘cause people don’t feel comfortable looking after her. Um, it’s just really difficult. But you know, my partner had to give up work because of no nursery would cater for children like, with my daughter’s condition. Um, so yeah, it has had a massive impact on us.

Participant 11, father of female child in early group

Even with the offer of a tailored care plan, parents were deterred by the fear of damage to the cast and/or injury to the child, and a lack of one-to-one care that they perceived necessary:

I keep thinking of was if she was to have an accident or, you know, something would happen to her cast or, you know, it might affect her legs not be so stable and keep them in the correct position. I just thought it’s not worth it.

Participant 1, mother of female child in early group

Rather, kin care, either from those with prior experience of caring for a child with DDH or from close relatives, was favoured by those engaged in full-time paid work:

Where mum dealt with me in a cast and things she knew what to do with my daughter in this same situation and I didn’t feel comfortable letting anyone else have her.

Participant 1, mother of female child in early group

... she can’t go to nursery ... well she can but personally I wouldn’t like to put her in a nursery. She’s hard work for me never mind somebody else taking care of her who isn’t used to the cast so now my mum and mother-in-law they both look after her so it’s kind of a swap and change.

Participant 2, mother of female child in intentionally delayed group

My mum actually is having him 2 days a week and one of my friends was having him 2 days a week up until sort of a few months ago and then he was going to nursery 1 day a week but he’s now going to a childminder 3 days a week and then my mum has him the other 2 days.

Participant 6, mother of female child in intentionally delayed group

... her mum has her a couple of days and then works for 3 days and she’s 2 days with her auntie and 1 day ... 1 day with my parents.

Participant 14, father of female child in early group

Some dual parent families were able to share and plan the care of the child around their work situations, although in all cases the mother was the primary carer. As noted above, those working shifts also negotiated home, work and family. This scenario, however, came with little respite as practical support from the wider family was not always forthcoming, with interviewees reporting the reluctance of relatives and friends to care for a child they saw as fragile:

... while she was like that so yeah everybody else was like ‘yeah I’d love to have her but, you know, we don’t want to because we don’t want to hurt her’. Or they weren’t sure how to pick her up or hold her or ... They all felt she was quite fragile.

Participant 1, mother of female child in early group

I think people are just very much like, no we’ll just leave that for you, you can, you know what you’re doing.

Participant 11, father of female child in early group
The perceived vulnerability of infants with DDH was also apparent in some instances within formal child care settings:

... they’ve kept her in the baby room at the nursery; she should be in the stage up, but I’ve asked them to keep her back ’cos I didn’t want, like, the older kids sort of trampling over her so she’s being kept in the baby room for the time being, until she’s a bit more mobile.

Participant 12, mother of female child in intentionally delayed group

For some parents, formal child care was the only option and settling their child proved to be an anxiety-inducing time:

Really anxious. I didn’t want to leave her ... I was just worried that they wouldn’t do things right because she was different from the other babies but they’ve been fantastic.

Participant 7, mother of female child in early group

One child attended a nursery where his mother was employed and, although she did not manage the room in which he was located, she was on hand to provide his carers with advice and guidance:

... they just asked me to talk it through to them what they needed ... like how to pick him up and stuff like that they were very happy and confident in like caring for him in the cast and stuff like that.

Participant 5, mother of male child in intentionally delayed group

Even in this case, the mother would have been reluctant to return to work had he not been cared for in the same building. That said, for those attending formal child care the support and specialist care was commended by parents:

... when we went to go and see the nurseries, um, you know, all of them were just like, no, it’s absolutely fine, we’ll just do a risk assessment there, and I’ve had a kid like that in here before. Um, you know, they were brilliant; I was quite surprised because I’d read quite a lot of negative things online ... people saying that wasn’t possible.

Participant 8, mother of female child in early group

In this instance, a member of staff at the nursery had previously cared for a child in a hip spica cast.

Child care was less of an issue for those on maternity/parental leave during rehabilitation following surgery. Child care needs were sometimes resolved by mothers delaying a return to paid employment on the basis that the child’s needs were too significant, as the following extracts demonstrate:

I’m sort of I’ve known the whole while my son has been in his cast that, you know, I couldn’t get a job because my son needed full-time care.

Participant 4, mother of male child in early group

... in the hospital when I was enquiring about it, if he can go into the nursery because I was planning to put him into the nursery; they said yes, but then I just, I felt sorry for him, and I just wanted to look after him myself.

Participant 13, mother of male child in intentionally delayed group

The reluctance to place a child in a hip spica into formal child care provision was, in part, tied to expectations around good parenting and, in particular, mothering.

The timing of surgery was, therefore, implicated in child care decision-making. Many parents extended their leave to accommodate their child’s treatment or relied on other family members. Electing for paid
external child care was rare as parents were not confident to leave their child with anyone inexperienced in cast care. The general reluctance to leave their child meant that parents, particularly mothers, had little respite.

**Costs to families** The additional financial costs to parents were also significant with funds required for hospital visits and stays, including travel, accommodation and subsistence in addition to expensive (replacement) baby/toddler equipment and furniture. The amount reported varied between families, with more items documented in the interviews than in the diaries. Distance to the hospital, along with the primary mode of transport used for day-to-day journeys and the child’s perceived needs, particularly in terms of comfort, were listed. With one parent on maternity/paternity leave, and with the differing responses of employers to requests for leave compelling some to take unpaid time away from work, these additional costs not only came at a time of reduced household income for many, but were also required over a short space of time:

... it was added money that we couldn’t really afford, because I wasn’t at work yet, but . . . we had to find a way, you know, I’m lucky we’ve got family – they helped.

Participant 9, mother of female child in early group

... you’re being told your daughter’s going to have surgery in 3 days and you need, your car seat’s not right, so you’ve just got, you have to spend £300 there, and then – money we didn’t ‘ave . . . you have to spend out like hundreds of pounds there and then to get it, and it is, it is difficult, and it does put a massive strain on you.

Participant 11, father of female child in early group

The total amount spent by parents varied, with figures noted in the diaries suggesting in excess of £200 to > £900, although the interviews often revealed other significant costs. This expenditure was generally necessary over a short space of time from the period leading up to surgery to shortly after the operation. Pushchairs, car seats and high chairs previously purchased often had to be replaced to accommodate the hip spica cast. Equipment necessary for the child’s comfort, such as a beanbag, was also purchased by the majority. Much of this was sourced from mainstream stores rather than specialist manufacturers (which were considerably more expensive). The ‘double nappy’ approach used by many parents to accommodate and protect the cast from moisture, along with more frequent changing also resulted in additional expenditure. Travel, especially for those living some distance from the hospital, and parking for clinic appointments, as well as accommodation and subsistence costs for hospitals stays, were also reported as significant expenses. Almost 60% of parents explicitly reported experiencing financial difficulties at some point in the process.

To fund these expenses, parents spoke of reducing either personal or household expenditure on leisure and clothing, and in one case food, borrowing money on credit cards and drawing on savings. Others received support from members of their extended families:

But we were quite fortunate . . . my . . . my partner’s parents they . . . at the very beginning when they found out this was happening they . . . um . . . paid for a lot of equipment for us . . . the car seat, pram, beanbag. Um . . . they really helped us out financially.

Participant 7, mother of female child in early group

Two explicitly stated that they were able to draw on state support in the form of Disability Living Allowance, the amount of which is subject to an assessment of the child’s need and is variable. Parents are eligible regardless of whether or not they are in work. Others had successfully applied for a Blue Badge to assist with parking. One mother had received support from a local charity (totalling £300) to cover the costs of travelling to and from hospital. A key DDH charity helped financially by offering expertly adapted car seats for loan. The uptake of state or charitable support was not common among the pilot families, with the exception of the car seat loan scheme.
Some parents spoke of the infuriation they felt by money wasted on inappropriate items. Participant 2, for instance, complained about the advice received in hospital suggesting that the brands recommended were not necessarily appropriate for all cast structures and that families should be advised to refrain from purchasing items until after surgery:

*I don’t think there’s a standard piece of equipment that you can buy for all children, you just need to wait and see what shape the cast is but brands-wise they [hospital staff] were very focused on each individual brand, which I don’t think was the best because actually one thing that we bought she didn’t fit.*

Participant 2, mother of female child in intentionally delayed group

*. . . a new car seat which she couldn’t sit in anyway after wasting money buying one.*

Participant 1, mother of female child in early group

Issues with the car seat were also implicated in her personal mobility while her child was in cast and she was reliant on, and constrained by, the high cost of local public transport.

Items suggested to participant 3, this time by a third-sector organisation, were not suitable for the shape of cast with which her daughter had been fitted:

*I did buy this other high chair that was recommended . . . but the surgeon likes to cast quite wide set but she still didn’t fit into it so she’s only been . . . fitted into that in the last 7 weeks [when] she’s been in broomsticks.*

Participant 3, mother of female child in early group

Two families used innovative solutions to overcome the challenges presented by a lack of practical, and indeed affordable, seating options for children in a wide hip spica cast, making their own spica chairs/table to ensure their children could participate in day-to-day activities, such as eating with the family.

It is apparent, then, that during the period before and immediately after surgery families encountered a number of additional costs including (1) travel and hospital stays, with the amount needed dependent on proximity to the hospital and/or nature of the journey, and (2) equipment based on the perceived needs of the child. Although some families were able to resource these costs without putting a strain on their finances, eight experienced difficulties at some point, relying on making cuts to their household expenditure or donations/loans from relatives. The state and third-sector organisations also provided support, but this does not appear to have been widely utilised.

**Familial relationships**

Caring for an infant undergoing surgical intervention for DDH did put a strain on relationships within the family.

**Couple relationships** We asked participants if they felt that their DDH experience had impacted on their relationship with their spouse/partner. For some, this was not a subject that was easy to discuss:

*I’ve struggled with a few questions at the end ‘cos it said about the sort of impact on yours and your partner’s relationship and I didn’t really know if that was applicable at all.*

Participant 7, mother of female child in early group

Of the 12 participants in couple relationships, half did not believe that it had affected their relationship in any way, with a further two interviewees feeling uncertain about the implications.
Three participants explicitly discussed the negative impact that their experiences had had on their relationship, as the following examples demonstrate:

. . . because I wasn’t able to get out um, you know, most days um . . . it just, yeah it was very wearing. It did make me feel quite low um . . . and, you know, that put strain on me and my husband . . . because obviously I felt like I wasn’t coping as much . . . because I wasn’t happy . . .

Participant 4, mother of male child in early group

I was going to say it did cause a bit more extra stress, we did argue a bit more than usual . . . I think that was due to being in the hospital for 2 weeks, we never actually got time together for 2 weeks . . . ‘Cause sometimes you focus so much on her, and nursing her, and helping her, that you sort of lose, you know . . . we lost a bit of a connection for a bit over this, ‘cos we were arguing, and then we were fine.

Participant 9, mother of female child in early group

. . . in terms of our relationship, it’s been quite strained at points. Um . . . but like . . . it’s, it’s weird; it’s hard to explain because it’s sort of brought us closer together; we’ve had to work together . . .

Participant 12, mother of female child in intentionally delayed group

Despite the challenges and strains encountered, the experience also proved bonding for some couples, as exemplified by descriptions of the emotional support partners provided for one another:

He’s been brilliant really although he actually started um . . . Googling [Google Inc., Mountain View, CA, USA] the condition [laughs] . . . and was telling me things and saying ‘oh it’s going to be all right’ and I was supporting him so um . . . yeah we were quite lucky, you know, we talked to each other about um . . . how we were feeling as well.

Participant 7, mother of female child in early group

We was support-, supportive of each other yeah. We kept each other going basically.

Participant 10, mother of female child in early group

I think I’ve just been a . . . a shoulder to cry on on the tough days, ‘cause obviously she’s the one at home with it with our daughter you’re, you know, all day every day, and I’ve . . . all I’ve done is just been a shoulder to cry on . . .

Participant 11, father of female child in early group

Sibling relationships Despite the body of work on the impact of sibling illness or disability on sisters and brothers, effects on siblings have yet to be acknowledged in the DDH literature.20 However, siblings can be highly influential in supporting their sisters and brothers. Five of the 14 parents had children older than the infant undergoing surgery for DDH. These children were each aware of, and affected by, their sibling’s surgical intervention. Even young children were aware of the absence of their sibling during hospitalisation:

She [2-year-old daughter] was definitely picking up on, that something wasn’t right obviously when my daughter was going into hospital for the treatment, like we were away for, like, 2 days with her. Um, and you . . . I get back and me, me mum had been looking after her and she said, ‘She’s not herself’. Like, she’d been ask–, she’d been asking for the baby.

Participant 11, father of female child in early group

It also had implications for sibling bonds:

I think what she found a little bit difficult is that um, she’s desperate to hold her little sister . . . but because she’s in a cast she’s really heavy and quite awkward, she hasn’t really been able to pick her up.

Participant 8, mother of female child in early group
Participant 5’s 5-year-old twins, for instance, had also been affected, she believed, not only by her and her youngest son’s absence from the household, but the delays to surgery, which led to a great deal of uncertainty:

They’d see me now and again and their brother. They were just missing their brother. And they knew what was happening because we spoke to them about it but they didn’t understand. Where we kept coming back home they were just getting really confused.

Participant 5, mother of male child in intentionally delayed group

Both became withdrawn socially around the time of surgery. Similarly, participant 6’s 9-year-old son had also been confused by the situation:

It’s been quite hard on the whole family. I’ve got an elder son as well who has been told his baby brother has, sort of, got to go in for an operation. He was, sort of, a bit distraught and he couldn’t quite understand why and what was wrong.

Participant 6, mother of male child in intentionally delayed group

Time was also an issue for parents as they felt they had less time to spend on their older children:

. . . he [brother] was sort of quite emotional and obviously we had less time to spend with him because we were obviously back and forth with my son to the hospital . . . which was obviously quite hard but um . . . but I’m quite lucky because he’s very understanding.

Participant 6, mother of male child in intentionally delayed group

Despite attempts to minimise the impact and shelter siblings from the details, anxiety was still an issue:

. . . she did worry, so I know that, like, when we went to take my daughter into hospital she was very distressed when we dropped her off in the morning; she was desperate to know, and for us to let the school know, um, so she would know, that obviously she’d come out of the operation and that it was fine.

Participant 8, mother of female child in early group

One mother felt that her older children had not been particularly affected but had offered considerable support:

They’ve been supportive as well, the ba-, the children. So, um, it was explained . . . as long as you explain to the children what you’ve been explained to, they’ll understand like us . . . so they’ve, they’ve helped out a little bit as well, the children.

Participant 10, mother of female child in early group

Parental physical health and mental well-being

This section outlines the wider effects of the condition and treatment on the health and well-being of parents and carers. For parents/carers the DDH journey can start long before formal diagnosis with a concern about the child’s physical development. As outlined in Experiences in primary care, of the 14 pilot parents, eight experienced some form of delay in gaining a formal diagnosis. For them, this part of the journey was rife with anxiety and stress as they battled for their concerns to be heard. Although the overwhelming majority provided accounts of what was often referred to as an ‘emotional rollercoaster’, for some their child’s condition and/or experience of treatment was regarded as detrimental to their own mental well-being:

I think I was quite emotional, um, and there were times when . . . I don’t know if it was when my partner had gone back to work, or if it was that initial, like, first few days at home, um, and I just looked at her at one point as if, like, I thought I don’t even know where to start with changing you . . . and I was quite close to sort of breaking down; um but I looked at her and she just smiled at me, and I was like, ohhh, it sort of snapped me back out of it to be fair.

Participant 12, mother of female child in intentionally delayed group
The following example illustrates the feelings of guilt that the mother, having had DDH herself, felt for her child’s situation:

I think . . . to start off with I sort of blamed myself and thought it was, you know, my fault for her having it and that made me quite upset and every time I looked at her I felt upset because I thought it was my fault but obviously I know it's not.

Participant 1, mother of female child in early group

Here, the legacy of her (and her mother’s) own experiences of DDH was implicated in her emotional well-being during her daughter’s journey through treatment. Family history, therefore, does not just relate risk factors for DDH but is also implicated in the way in which parents cope with their own child’s journey.

After experiencing the repeated dismissal of her concerns about her daughter’s hip by both her GP and her local hospital, one new mother spoke candidly about the connection between her daughter’s condition, her anxieties and the onset of postnatal depression, for which she was receiving treatment:

I have up and down days . . . most days I’m really fine and coping well. I start to get a bit anxious before we go back into hospital . . . cos I don’t like the whole hospital experience which really just put me off . . . just everything that’s happened . . . [becoming tearful again] . . . and also the worry that the hip might have regressed in the cast. She might have hurt it. It might be dislocated. I was also quite happy, positive before but [triggered] a bit of PND [postnatal depression] now.

Participant 3, mother of female child in early group

Transporting a child in cast and attending baby groups proved problematic, if not impossible, leaving some new mothers in danger of feeling isolated:

I felt quite unsociable in a way and a little bit isolated.

Participant 1, mother of female child in early group

Indeed, recent research suggests that a significant connection between employment status and psychological well-being, with those outside the labour market, including full-time carers, faring worse. One mother had previously experienced both depression and agoraphobia, and was conscious that social isolation brought about by being unable to easily transport her daughter by car could adversely affect her own mental well-being if she did not push herself out of isolation:

I mean I’ve suffered with depression . . . on and off and um my concern was sort of how I was going to cope on that front . . . and I also had agoraphobia as a child . . . um so I knew that by staying in and not going out all of the time it could potentially bring my anxiety back because I could quite happily kinda get back into the routine . . . of not wanting to go out. So I was aware of that happening if I wasn’t careful . . . It did make me feel quite low um . . . and, you know, that put strain on me and my husband . . .

Participant 4, mother of male child in early group

The effects on mental well-being were, for some, temporal, featuring during particular stages of the journey. One mother, for instance, spoke of exhaustion through sleep deprivation during the period following each cast change, and from undertaking long commutes between work and the hospital for cast repairs:

Probably worrying myself makes me feel exhausted but her waking up and just the um . . . the trips back and forth to the hospital because the first cast she had on . . . she’s in the second cast now . . . but the first one it kept sort of . . . and it looked like it was breaking so we were back and forth to A&E [accident and emergency] at night after work um . . . which was just really tiring and then having to be awake with her in the night and then back to work again in the day. It was a tough couple of months.

Participant 7, mother of female child in early group
RESULTS

This example also highlights issues of timing. Her daughter was aged 13 months at the time of surgery (early-treatment arm). She had just started a new job and it was evident in her interview that her levels of stress were exacerbated by having to juggle work, a long commute and her daughter’s additional care needs as highlighted in the quotation, and having to ask a new employer for leave.

Even for those who did not explicitly discuss mental health issues, either stress or sadness often featured in their accounts:

Um . . . [sighs], yeah I think it was just, you know, [sighs], we tried to be really positive, but there are inevitably those moments when you feel a bit hard done by, and it sounds silly but for me it was like we went to a clothes shop with her, and you’d be like, she can’t have anything in here . . .

Participant 8, mother of female child in early group

There was loads . . . there has been loads of stress.

Participant 11, father of female child in early group

. . . just the apprehension about the unknown so how to deal with it when it actually happens . . . because you’re not in control and because um . . . they need to go through the surgical procedure so there’s that worry.

Participant 14, father of female child in early group

Four participants, all of whom had children aged > 11 months at the time of surgery, believed that their child’s condition had had an impact on their physical health. For some, issues related to handling their child and the weight and awkwardness of the cast resulted in parents suffering shoulder and back pain:

She’s very heavy in the cast now which we weren’t expecting to be honest . . . for her to be that heavy.

Participant 2, mother of female child in intentionally delayed group

. . . the only thing that I think that’s it’s caused is problems with my partner and mine’s backs . . . The small of our backs do hurt because you find yourself leaning back a bit to sort of compensate really for holding and er yeah we’ve both got sore backs.

Participant 3, mother of female child in early group

. . . she’s just wriggling around and it’s just the fact that she’s so much heavier and awkward so kind of lifting her in and out [of the car seat] . . . takes a bit more time and it pulls on your back a bit and that’s probably the only main difference it’s just really a lot more awkward and heavy for us.

Participant 7, mother of female child in early group

I would just say like, um, back and shoulder pain . . . from lifting her . . . um, but you know.

Participant 12, mother of female child in intentionally delayed group

There was also concern for the physical health of grandparent carers:

I do [work] 4 days a week now I’ve gone back, um, and she does 2 days nursery, 1 day at grandma, 1 day with nana – I think our concern was almost more about how would the grandparents feel with how to pick up, what is . . . what is, you know a heavy child in awkward positions, and whether their physical health was up to that.

Participant 8, mother of female child in early group

. . . if people looked after her for a long period of time in the spica and were carrying her then they would . . . they would get a bit more fatigued than normal.

Participant 14, father of female child in early group
Handling the child in public spaces was especially difficult, as will be discussed further in the following section.

**Family mobility and isolation during rehabilitation**

One key issue for families was the transportability of a child in cast. The ease at which parents could use existing or readily available car seats and prams/pushchairs was dependent on the shape and width of the cast. Despite the guidance received from occupational therapists at the sites, or from the aforementioned national charity Steps, from whom a specially adapted car seat could be hired, family mobility and isolation during rehabilitation proved to be an issue for nearly three-quarters of the pilot families.

Four parents had not experienced any restrictions to their mobility while their child was in a cast. Three of these mothers had older children and were employed in full-time occupations, and it is unlikely that they would have had any alternative but to maintain the family’s daily routine. The fourth mother was a full-time carer and lone parent who travelled mainly on foot.

The shape and nature of the cast placed some short-term restrictions on the mobility of some families, particularly car travel as exemplified by the following examples:

*We could’t take her out in the car so now that we can take her out in the car which is nice. We’ve been able to visit people that, you know, have always had to come and see us.*

**Participant 1, mother of female child in early group**

*... getting out and about is quite difficult because of getting her in the car and the weight of her it’s just quite tricky.*

**Participant 2, mother of female child in intentionally delayed group**

*In the second spica cast the shape of that was so awkward that there were a few occasions I couldn’t get her done up in that car seat. My partner really struggled so there was a couple of times I think I meant to go out and I just be [trying] for 15 minutes and I couldn’t get her in it ... I think for a while I stopped going to playgroups and stuff but I have started back up again.*

**Participant 3, mother of female child in early group**

The difficulties these mothers faced in securing their children into a car seat prevented them from travelling by car and stopped them attending baby-oriented activities outside their immediate vicinity. This was often a relatively short-term issue that was resolved either by the purchase or hire of a new car seat:

*It was tricky for the first time we went out into the car, but after that we just got ‘er in ... it just wasn’t as bad as we thought we’d be, we managed absolutely fine.*

**Participant 10, mother of female child in early group**

*... we were housebound for a couple, I think it was like a few days, um, ... which was I suppose fine because I don’t know that we could have gone out at that point, but it just, it felt like a bit more restrictive because we didn’t have the stuff and couldn’t go out.*

**Participant 12, mother of female child in intentionally delayed group**

For others, the ease of portability of the child had a longer-term impact on the family’s mobility. This was more prevalent for those reliant on car than those used to travelling on foot or by public transport. For parents with other children, day-to-day tasks, such as shopping with one child in a cast, also proved challenging:

*We can’t all go food shopping together because um, ‘cause obviously by now we just put ‘em [both children] in a double er trolley you know, the ... you know, we just put ‘em in the double one of them, but now older daughter has to go in the trolley and daughter with DDH has to be pushed in the pram.*

**Participant 11, father of female child in early group**
The managing of care tasks in public was also an issue that confined some parents to the home/relative’s homes or restricted the duration of journeys and activities as one mother described:

*I was conscious that I would go out but plan to be back to change her [nappy] . . . so I’d nip into town, nip into the supermarket. Um, the furthest I’d go might be to my parents ‘cause I could change her there . . . but wouldn’t sort of go out for the day with her.*

*Participant 12, mother of female child in intentionally delayed group*

This mother described missing out on baby-oriented activities while her daughter was in a cast, and by the time the cast had been removed she had returned to (part-time) employment. She was also concerned about suitability and safety of certain activities:

*I did go to a couple of um, er, mummy and baby groups . . . but you couldn’t actually, obviously, do a lot at all. But it became a little bit . . . it does get a bit frustrating ‘cause I suppose she’s just laid there and I did feel sorry for her.*

*Participant 9, mother of female child in early group*

Indeed, as previously mentioned, the fragility of the child was alluded to in many interviews. Short-term restrictions on the mobility of the parent and child were also resolved through forward planning and adaptation to both the activities in which they engaged, and the equipment used:

* . . . it’s more about just adapting her to those situations . . . So there’s been a lot more planning . . . and it’s been a lot more difficult when we’re out just to make sure she’s comfortable and to lift her and so on.*

*Participant 7, mother of female child in early group*

For some, exclusion from such activities had wider implications for the parent–child bond:

*We can’t just go to the park. She can’t do normal things that kids would do to bond with us so, yeah it has been quite difficult. [Before] We done quite a lot with her. She’d been out all the time. She’d always be in the park. She was a very cuddly baby . . . and now obviously she can’t do that so yeah it has been a big change for her.*

*Participant 2, mother of female child in intentionally delayed group*

For others, isolation was a long-term feature of their DDH journey, which affected their own well-being, as one mother described:

*I think the hardest thing um . . . has been that he didn’t fit in his car seat . . . oh originally he did but it was very . . . we needed two of us to be able to push the seatbelt so hard to get it to sort of click into place . . . so I was um stuck at home pretty for much for the duration of the treatment which, for me, I found emotionally quite difficult . . .*

*Participant 4, mother of male child in early group*

The isolation experienced by some parents could have longer-term impacts on their ability to develop family social capital, defined as resources individuals and collectives derive from their social networks that are essential for resilience during challenging periods.21
The ability to be able to transport a child in a cast was vital to the functioning of some families, especially when the children lived between two homes or when day-to-day care was multisited. One father spoke of the potential impact of not being able to transport his daughter on the working and care lives of his extended family:

*Her mother and I are in separate houses. We’re separated. So there was that need to move between . . . it’s one of those things where you understand [the logic behind the cast shape] and can support that . . . but also you need to um . . . be able to live for the next 6 weeks.*

Participant 14, father of female child in early group

Although not a specific focus of the interview, the use of the Blue Badge scheme that enables drivers or passengers with disabilities to park closer to their destination, was mentioned by two parents as an asset to family mobility.

**Participant feedback on involvement in the trial**

The final section of this report details the feedback provided by parents about their motivations for, and experiences of, taking part in the trial.

**Motivations**

Participants described a range of commonly cited motivations for taking part in the trial, with benevolence towards others and the advancement of medical knowledge key factors for the majority:

*The term ‘conditional altruism’ concisely describes the willingness to help others that may initially incline people to participate in a trial, but that is unlikely to lead to trial participation in practice unless people also recognise that participation will benefit them personally.*

McCann et al. 22

Similarly, eight parents were driven by a desire for research that would help ensure that their own future children, and in one instance grandchildren, or other (unknown) children would not have to endure the same experience. For some, this was couched in terms of further research having potential benefits for the welfare of children and families, to ease suffering, as the following examples highlight:

* . . . to find out why children are born with this condition and how it can be prevented so others do not have to suffer the heartache my family have.*

Participant 6, mother of male child in intentionally delayed group

*I’d like to take part in this to help any future, you know, parents or guardians that are going through this . . . because it has . . . it has been quite worry and if there’s anything I can do to kind of help . . . um research this on in the future then that’s brilliant.*

Participant 7, mother of female child in early group

*. . . because it would be helpful for the other parents to know what it is like . . . and they don’t have to go through what I went through, or the parents before me . . .*  

Participant 13, mother of male child in intentionally delayed group

This was especially the case for parents with a family history of DDH, as one mother stated:

*Mainly because I think where I had it and she’s got it and if we have any more children in the future that obviously they’ve got a chance of getting it as well then, you know, in years to come when they have children then it could happen again. So I thought at least for us taking part will help in a few years to come, you know, what is the best outcome.*

Participant 1, mother of female child in early group
For others, the impetus centred on supporting fellow parents, particularly in terms of reducing the burden of responsibility to make the right decisions about their child’s treatment:

> Hopefully other parents won’t have to make that decision because they’ll know what is the right way forward.

*Participant 8, mother of female child in early group*

For a small number of participants from different research sites, the motivation to take part focused primarily on the treatment of their own child, with three suggesting that participation in the trial would result in better-quality care or swifter treatment:

> . . . if I had gone through it without being on the research I wonder whether I would have had as much, sort of, advice . . . as I have done.

*Participant 6, mother of male child in intentionally delayed group*

> I spoke to a few of the nurses and the surgeon and it just seemed like the best care, and they were putting her like first more, you know, instead of waiting and going through the normal way they seemed very professional, . . . I suppose; it seemed like she would get more out of it.

*Participant 9, mother of female child in early group*

> . . . if we didn’t get put into this scheme [trial] we would have to wait years for daughter to have surgery.

*Participant 10, mother of female child in early group*

> . . . truthfully, the reason was we got told there’s a chance we could have got the surgery quicker.

*Participant 11, father of female child in early group*

The emphasis on patient’s participation in randomised controlled trials as driven by altruistic tendencies as well as perceived personal benefits has been documented by others.22

Although in their demographic questionnaire all interviewees stated that they felt well informed about the trial and had sufficient time to discuss the surgical options, participants from two of the four sites had not viewed the informational film. Nonetheless, the motivations for trial participation cited suggested that some were unclear as to the purpose and potential outcomes of the research, with six inferring that the trial would shed light on causality, rather than the efficacy of the timing of surgery. Indeed, there were only three examples when an explicit reference to the temporal focus was mentioned, all of whom had studied the informational film:

> . . . we did a bit of research. We know there’s not a lot known about when is the best time to operate and um . . . where it’s . . . where it’s such a strong family history, you know, there might be a chance that our second child then has this condition . . .

*Participant 3, mother of female child in early group*

> . . . it just all felt quite confusing with, you know, what was a success and what wasn’t and what route to go down whether for him to have it early or whether to have it late done you know actually obviously it would be more helpful for people to know when is the right time.

*Participant 4, mother of male child in early group*

> . . . after looking into it all, I don’t really know what option we would have taken. So . . . but when there doesn’t seem a right answer.

*Participant 8, mother of female child in early group*
Levels of understanding cross-cut the diversity of the sample with those demonstrating a clearer understanding of the trial’s purpose having a variety of educational and occupational backgrounds. Indeed, during the interview some described feeling confused about the trial:

... they sort of said it was your choice, but when they were talking through it, it just, it didn’t really make sense what they were saying. Um, but deep down it felt right to take part in the trial.

Participant 12, mother of female child in intentionally delayed group

English was an additional language for the only parent who had viewed the informational film but had not found it helpful.

The feedback garnered from the pilot interviews may help shed light on the recruitment challenges faced by the trial. Many of the interviewees were driven by what they saw as the study’s potential to make the processes for surgical treatment more definitive, reduce the decision-making burden of responsibility on parents and understand causality, suggesting that the trial’s remit was not fully understood.

Perceptions of randomisation

Nine of the 14 infants of interviewees were allocated to the early-treatment arm, whereas the surgery for the remaining five was intentionally delayed. As outlined in the previous paragraph, helping future children was one of the key motivating factors in trial participation. That said, the interview accounts often highlighted the burden of responsibility and anxiety felt by many over consenting to take part, as summarised by participant 8:

I think we felt if we were gonna to sign up to the trial we had to be prepared for either option, and to be equally comfortable really with both, both, both groups – does that make sense?

Participant 8, mother of female child in early group

Within the clinical literature, the timing of surgery is currently equivocal. However, some of the pilot interviews expressed a clear preference based on their understanding of the information and advice received. Although some voiced anxiety around the uncertainty resulting from randomisation, allocation to their preferred option was met with relief:

... we were concerned as to which um ... one ... when he was going to have it done. Yeah, yeah. And I think initially we actually wanted it ... we were hoping for it to be later ... which actually I’m glad we didn’t [laughs].

Participant 4, mother of male child in early group

... fortunately it [randomisation] did go for the closed surgery um ... when they selected the option for us, which was our preferred choice.

Participant 14, father of female child in early group

Despite agreeing to take part in the trial, for some the outcome of randomisation would have affected the likelihood of their remaining on the project:

The consultant told us we’d either get it quicker than usual, or later than usual, and if we’d have backed away until she was 1 we wouldn’t ‘ave, we wouldn’t ‘ave gone through with it.

Participant 11, father of female child in early group

... only if we’d perhaps selected the open surgery. I think we may have then wanted to have more conversations and more talk about that ... the way it happened then no.

Participant 14, father of female child in early group
For one participant, the option of withdrawal should an unfavourable treatment arm be allocated was alluded to by research staff:

*There’s no harm and she said if you don’t like it you can always pull yourself out. I said ‘Well I might as well do that if I can . . .’. If I don’t like it then all I have to say is ‘No I don’t want to do it’ and then I can go back to the normal route.*

Participant 5, mother of male child in intentionally delayed group

Furthermore, the confusion expressed by some participants over the purpose of the trial also extended to understandings of the process of randomisation. The examples cited here, from different sites but both allocated to the intentionally delayed arm, imply that some felt inadequately informed or misled:

* . . . two nurses in at the time talking to us about the trial, um, but myself and my partner came away with different views in terms of what they were actually saying, um . . . and in terms of they were talking about the different treatments that open and closed . . . and that if you went on the trial you could get one, but you couldn’t get the other.*

Participant 12, mother of female child in intentionally delayed group

* . . . I’ve heard it from the, or from the surgeon who was operating on my son . . . that if, that I would like to have the earlier . . . but then afterwards the lady, they did, er, the surgeon from the trial told me . . . because I went on the trial, so they can decide for me . . . And this no one told me so I thought like, that it was kind of misleading me.*

Participant 13, mother of male child in intentionally delayed group

One parent even expressed some cynicism about the underlying rationale for the trial:

* . . . alarm bells just rang when they mentioned the trial, which is probably silly, but . . . I just thought, are they just trying to find like the slightest thing and then getting them in for the operation], rather than seeing if other options will work, or if it will just correct itself.*

Participant 12, mother of female child in intentionally delayed group

Confusion, even cynicism, over randomisation in randomised controlled trials is not uncommon. Once participants had made the decision to take part in the trial, the majority did not have any further worries or concerns about the research.

With a young child and already much uncertainty around the outcomes of surgery it is, perhaps, to be expected that many felt concerned leaving the path of the child’s treatment to a process referred to as ‘randomisation’; a process that has otherwise been perceived of as haphazard.

**Experiences**

During the interview, parents’ views on their experiences of taking part in different aspects of the trial were garnered. Although talking candidly, but not anonymously, to a member of the research team is likely to have shaped the responses and levels of disclosure, the distinct situation of the qualitative team, part of but at the same time separate from the main trial, was made clear to participants. As the following subsections focusing on different elements of data collection show, many were forthright in the evaluations of their experiences.

**Clinic** The clinic visits, as an aspect of data collection, were generally experienced positively, with many making explicit reference to the advantages of attending a dedicated research clinic. Based on both actual experience and perceptions of other clinics, some parents believed that they had been afforded more time
and attention in a quieter, less stressful setting, where there was the potential to build longer-term relationships with particular health professionals, as the following examples illustrate:

I feel we’ve been looked after a lot more actually than what we would have been if we hadn’t been part of the trial. I think a lot more focus has been on us.

Participant 2, mother of female child in intentionally delayed group

The research clinic has been a really nice place to go to because it’s a lot quieter than um . . . the main clinic . . . and the people, you know, it’s nice to see the same people . . . that’s what we’ve really appreciated as well um . . . and to gain that relationship with those people.

Participant 4, mother of male child in early group

I’m not saying you’d be treated like a number but it would be like ‘Oh this, this and this sort of thing’ but when . . . but now looking back like all the nurses knew him and knew me ‘cos the research team . . . the research team would come up and greet me and stuff like that . . . so I kind of felt like . . . I kind of knew my place because they would talk to the nurses on board. The consultants would then say I’m the researcher then they’d spend more time getting information from me. In that aspect of it I would get more information out of them . . . from it.

Participant 5, mother of male child in intentionally delayed group

Yeah, I mean, I think that, you know, that’s all been fine really. I mean obviously, you know, one of the small benefits is that, you know, the, the hip clinic is obviously . . . it’s a calmer process.

Participant 8, mother of female child in early group

. . . it was really enjoyable, and very nice, because as soon as we went in there for our appointment she was straight in . . . there was no waiting, you know . . . it was . . . they were all friendly, they’re all . . . it was just, . . . I was so glad that I did it, that I did it, you know, that way.

Participant 9, mother of female child in early group

. . . we had only been into the other [mainstream] clinic once – that was the first time, that was when he [surgeon] asked us through and, like, the nappy was hanging off and all the rest of it. Um, but it just, I don’t know, it just feels like they’ve got a little bit more time for you.

Participant 12, mother of female child in intentionally delayed group

The attention and additional level of care that parents perceived to have been afforded by research staff ultimately aided their own personal journeys through the process, helping them to feel more at ease and supported. Indeed, the demographic questionnaire and the interview were both used, by some, as a means of communicating appreciation and affording praise to particular (named) staff, especially consultant orthopaedic surgeons.

Criticisms again included a lack of clear information:

. . . we did have some quite differing information . . . so I think I felt quite confused at one point; I think we went for three different appointments and got three different stories . . . um, and it was just very difficult to understand exactly what was going to be happening, so I, I would . . . just say maybe just a slightly more consistent approach.

Participant 7, mother of female child in early group

. . . we got told that we were gonna ‘ave more appointments with the surgeon which hasn’t ‘appened, er, which isn’t a problem, but . . . we were told that she was gonna get checked on more by him.

Participant 11, father of female child in early group
Despite some of the aforementioned concerns about the level and clarity of information imparted to the parents of trial patients, the experience of attending a research clinic for assessments was generally positive.

**Questionnaires** For the majority of participants, the questionnaires (including the demographic, CarerQoL questionnaire, Oucher Pain Scale and PedsQL inventory) were considered simple and straightforward. Unlike other aspects of the trial, the questionnaires were neither a burden on time nor anxiety-including, as summarised by the following example:

> I think generally you fill out a bit of paperwork when we’re there but generally it’s only a couple of minutes before we go in . . .

*Participant 14, father of female child in early group*

That said, the purpose of some lines of questioning was not always clear:

> . . . sometimes the questionnaires and stuff like that they ask questions but they’re not very specific . . . in what they say and that’s what I get really confused at . . .

*Participant 5, mother of male child in intentionally delayed group*

In one site, help from research staff was explicitly mentioned:

> The nurse helped us out with that [questionnaire], so . . . ‘cos a lot of things I don’t understand much of what it’s saying.

*Participant 10, mother of female child in early group*

There was a suggestion that the questionnaires were too focused on particular aspects of the experience and did not capture the broader impacts:

> . . . the questionnaires, um, I feel that they are mainly about the financial side. Not how, about the practical things, about the kids . . . that maybe they should put more about day-to-day tasks.

*Participant 13, mother of male child in intentionally delayed group*

**Diaries** The parental cost diaries were designed to garner data on primary and community care resource use, medications prescribed and purchased, the costs to families accrued through, for example, hospital visits and stays, and the purchase of additional equipment. It was intended that new diaries would be distributed at each visit and completed entries collected. Although the questionnaires were well received, this method of data collection attracted far more criticism from parents. Completing, and remembering to include information about costs, proved burdensome for some during a time when their child was undergoing treatment or when they were undertaking additional care tasks particular to a child in a hip spica cast. Participant 9 outlined her feelings:

> It was quite stressful trying to keep up with this diary when we were trying to sort her out, and you know . . . and sort out . . . the family life and everything getting, getting everything together . . . so I found it a little bit of a pain almost . . . like jot it all down ‘cause there are certain that you forget or that you don’t really remember properly because it’s quite stressful . . .

*Participant 9, mother of female child in early group*

Given the cessation of the trial, the intended analysis of the diaries was not possible. Nonetheless, it has been fruitful to cross-analyse the diary entries with the interview data. As demonstrated in *Finances and resources*, the interviews proved essential to understanding expenditure and providing supplementary data that some omitted, either intentionally or accidentally, from their diary entries.

> . . . it has been hard to remember to fill in the diary . . . particularly the . . . like um if you’re giving them any medication . . . and I think actually it might be partly as to how it’s been put because I think
it would be better if it was more like a tally . . . kind of way of filling it in . . . rather than it being how many times during the week or whatever . . . because that’s a lot harder to sort of keep on top of . . . whereas I think if it was just a tally you’d feel like it’s a lot easier to stay on track with it.

Participant 4, mother of male child in early group

I’m used to doing lots of paperwork [laughs] anyway so that doesn’t take . . . doesn’t take much time . . . Obviously I sort of . . . I tend to forget to do it as it happens . . . so I’m then thinking back and ‘oh I’ve forgotten this’.

Participant 5, mother of male child in intentionally delayed group

One further issue related to the operationalisation of this method by the sites. During the interviews, some complained that the diaries did not provide enough space for parents to expand on the reasoning behind certain purchases or to justify the inclusion of particular expenses, as articulated by participant 1:

The log book [sic] that we fill in probably could do with . . . like a few note boxes so you could er . . . like extend on certain parts of ‘why’ or the reasoning behind it ‘cos on a couple I’ve filled in I’ve sort of wrote a bit extra on it.

Participant 1, mother of female child in early group

Some were not clear as to the nature of the expenses that needed to be included. For others, the process of submitting entries and receiving new diaries was also unclear:

I think I spoke to her [research nurse] while I was in the hospital and said, Oh I need another one, um, and she didn’t send it out; and then I e-mailed her and she didn’t send it out, so I thought, Oh maybe, like, at this point you don’t need it . . . Um, and then when we went for our next er, first cast change in January, she then said: Oh, um, can you fill this out, or can you do this . . . and I was like, Oh, you know, it’s like I’ve been chasing, and actually I don’t think it was even January, I think it was like the next appointment in March, when she was coming out with it, um she turned up with a pack and asked me to fill it out.

Participant 12, mother of female child in intentionally delayed group

The challenges associated with the use of diaries in health and social research has been widely acknowledged.25 For the parents in this pilot study, these challenges were likely to have been exacerbated by (1) their experiences of having a young baby undergoing testing and interventions for DDH, (2) the timing of surgery as coinciding with the processing of returning to/find new employment after a period of maternity/parental leave, and (3) a lack of clear and consistent instruction on the completion and submission of diary entries.

Interviews Providing feedback about the telephone interview during the conversation was not necessarily conducive to candid disclosure. Although the duration of an interview is, by no means, an indicator of engagement or willingness to divulge, the average discussion, just under 1 hour, provided rich accounts of parent’s experiences from diagnosis through to aftercare and life beyond the cast.

Feedback garnered pointed to a positive experience, which for some was conducive to the expression of emotion despite the physical distance between the researcher and participant as exemplified by participant 5:

I was very confident in my answers and the interview on the phone I’m pretty happy.

Participant 5, mother of male child in intentionally delayed group

Other feedback was more implicit in that it focused on the shortcomings of other methods used, as outlined above, that did not provide the space to share lived experiences of treatment and rehabilitation, but rather focused on predetermined impacts on, for example, financial costs. Indeed, remote interviewing
can encourage the disclosure of sensitive and emotive subjects removing the pressure of the presence of the researcher, as stated by participant 3 when reflecting on the emotional account she provided:

*I knew I end up crying when I spoke about it so . . . It is . . . and when I talk about it, especially the first time to somebody, that’s when I tend to get a bit . . . I suppose I didn’t really know what to expect . . . but um it’s been fine.*

*Participant 3, mother of female child in early group*

For some, the interview was the preferred mode of participation, offering greater flexibility and convenience for family life, the demands on which are likely to be exacerbated by DDH treatment, as expressed by participant 9:

*I’d probably just say that, like what we’re doing now [telephone interview] is a lot more easier than telling the mums to write a diary out, you know . . . I think I would definitely recommend doing it this way, than giving them a diary each 6 weeks, or however they did it, because it just, it made more . . . I think it just put a bit more stress on it.*

*Participant 9, mother of female child in early group*

**Health economic aspects**

No worthwhile results could be taken from this aspect of the study.
Chapter 4 Discussion

Which timing of surgery is superior in the treatment of DDH – early or intentionally delayed? The Hip ‘Op trial was an ambitious trial that intended to deliver definitive, practice-changing evidence that could have not only given more certainty to future DDH infants and their families, but could have also contributed to the reconciliation of opinion within a divided paediatric surgical community.

The challenges of delivering the Hip ‘Op trial were recognised during trial development; consequently, an 18-month pilot phase and 6-month closedown plan to be activated should the pilot not meet its progression criteria were pre-agreed and written into the study design. Unfortunately, initial concerns were realised when the trial failed to recruit to time and target and closed early, in agreement with the study funder, after <5% of the total recruitment target had been reached. The main findings of the Hip ‘Op trial thus constitute a description of the challenges faced and lessons learnt.

Despite the disappointing outcome of the trial as a whole, some very valuable insight has been gained into the qualitative aspects of the DDH experience for the parents and wider families of the affected infants. The qualitative aspects are discussed in more detail in Qualitative aspects; however, the qualitative work initiated in the Hip ‘Op trial has provided the springboard for future research, publications and, ultimately, potential improvement in clinical practice and advice. Importantly, some of the very ‘real-life’ concerns surrounding DDH treatment, as highlighted by the qualitative work, were implicated in the trial’s difficulty to recruit.

Main findings

Study conduct

At the time of trial closure, 15 centres were participating, which is two more than were originally planned. Of these centres, 10 were open within the first 4 months. Most of the centres that opened after the initial 4-month period were delayed because the approvals process was much slower or more problematic than anticipated. However, at an overall study level, poor recruitment was not related to a lack of participating centres or delay in opening of centres to recruitment.

When the Hip ‘Op trial was designed, significant feasibility work was carried out to assess the willingness of both surgeons and parents to recruit children into this trial. A questionnaire was sent to surgeons, who were members of the British Society for Children’s Orthopaedic Surgery, and the response from leading centres indicated willingness to recruit patients to this trial. In addition, parents of children with DDH were asked to complete a questionnaire via the Steps website. The responses were favourable, with the majority being interested in their child taking part in the study and being open to randomisation.

All of the original proposed recruiting sites (13 in total) were contacted and asked how many patients aged <15 months were treated surgically for DDH: the total was 210 annually. These data are not routinely collected in, for example, the Hospital Episode Statistics system. Surgeons had to consult their local records and notes. Clinical experience estimated that about 20% of this population would present with a pre-existing ON and, thus, be ineligible for the trial. As such, it was concluded that approximately 168 patients would have been available annually across the centres surveyed. By the site contract stage of the approvals process, most of the proposed recruiting sites agreed to recruitment targets that were approximately 50% of the numbers provided initially. Interestingly, during the visits to individual sites, many of the investigators suggested that they had probably overestimated the numbers of DDH cases reported during feasibility. Some centres suggested that fewer patients were presenting late. In addition, it was suggested that referral patterns had changed in the 2 years between the initial survey and being asked to commit to a recruitment target; with children being more likely to be referred to a specialist centre with higher success in managing the condition with a harness, and hence avoiding the need for surgery.

© Queen’s Printer and Controller of HMSO 2017. This work was produced by Williams et al. under the terms of a commissioning contract issued by the Secretary of State for Health. This issue may be freely reproduced for the purposes of private research and study and extracts (or indeed, the full report) may be included in professional journals provided that suitable acknowledgement is made and the reproduction is not associated with any form of advertising. Applications for commercial reproduction should be addressed to: NIHR Journals Library, National Institute for Health Research, Evaluation, Trials and Studies Coordinating Centre, Alpha House, University of Southampton Science Park, Southampton SO16 7NS, UK.
The Hip ‘Op trial screening logs recorded a total of 118 children. This number does not include those children treated in a Pavlik harness; thus, it should essentially represent the population of children requiring surgical treatment for DDH. Twenty-eight of those children screened (approximately 24%) were reported to have been ineligible because of an existing ON, and a total of 74 children were eligible. The original feasibility study suggested 168 eligible children, whereas in reality fewer than half this number \((n = 74)\) were reported as eligible, and this is despite taking into consideration the fact that the actual screening included two more centres (15 vs. 13) and 4 months’ more recruitment time (16 months vs. 1 year) than the original feasibility work. Centres commonly reported that a substantial number of children were treated successfully in a Pavlik harness; however, most of the centres also said that they had not seen a significant increase in the rate of successful harness treatment in recent years. As such, unless the original numbers provided at feasibility were extremely inaccurate (in the order of about 50%), it would appear that the screening activity reported was not representative of the true patient population.

Interestingly, screening activity was not comparable across all sites; there was a large variability in screening activity between sites. It is possible that such variability could be accounted for by several factors (e.g. length of time open to recruitment, size of the centre, whether or not the centre is a specialist referral centre). However, in many cases the screening activity did not correlate logically with the length of time open to recruitment (or the size of the centre, etc.), thus strengthening the notion that screening activity was not reported accurately. The proportion of children reported as having an existing ON was also very variable between sites; however, in many cases, this was likely to be mainly as result of the inconsistent completion of the screening logs and the variability in the investigator’s choice of imaging technique and timing for ON detection, which we discuss later in this section.

The CI interviewed several of the site leads informally. Many expressed challenges to participation as a result of an inability to access local research infrastructure. Although the clinical trials unit and other investigators could tell local investigators what facilities should be available to them, the reality was that many were paediatric orthopaedic surgeons who were not routinely involved with research; they were therefore research naive and took a long time to navigate their way through local processes.

The pragmatic design of the study (e.g. clinician choice to use ultrasound or radiography, timing of early treatment as per local standard practice) seems to have allowed a certain degree of surgeon-/centre-level interpretation of various key aspects. Such variation in interpretation could also explain some of the intercentre differences in screening results and eligibility. For example, ultrasound is more sensitive than radiography when detecting the ON; as such, some centres cited their use of ultrasound as one reason why they had more ineligible children with an existing ON. Another centre stated that its standard practice was to wait until 5–6 months of age before considering surgery; thus, more children would have developed an ON by this age. Other centres suggested that they had fewer eligible children because they routinely left the Pavlik harness on beyond 12 weeks of age; thus, at this age, whether or not the harness failed, a child was ineligible for the Hip ‘Op trial. It is worth noting that the details of the study were already established at the time of the original feasibility and invitation to participate – centres whose usual practice differed from that described in the application had the opportunity to raise this with the project team, but signed up to the study without identifying these potential challenges to delivery.

Recruitment activity was also very variable between sites: as with the screening activity, in many cases recruitment did not correlate with logical factors such as length of time open to recruitment or the size of the centre. Of the 15 centres that were open, only seven recruited to the study: Southampton (open 16 months) and Alder Hey (open 14 months) recruited most patients, contributing 70% of total recruitment between them. In contrast, other centres failed to recruit any patients, for example, Barts and Sheffield (both open 15 months). Ten centres were open by the end of January 2015; by February 2015 recruitment was already below target and fewer children than expected had been recorded on the screening logs. This observation continued without improvement and despite the best efforts of the trial management team (see Appendix 3).
Of the eligible patients recorded on the screening logs, nine parents said that they did not want to take part in a research study and 17 said that they did not want the surgery to be dictated by randomisation. These two categories represent the families who might have agreed for their child to enter the study had the study been introduced differently, perhaps in a more balanced manner. Interestingly, one centre commented that, when informing families and answering queries about treatment, it felt that it could not state certain things because these things had not been the team’s own experience (e.g. one team felt that they could not tell families that further surgery was more likely if the primary surgery is performed early because this had not been their own team’s experience). Furthermore, a question that was asked very commonly by families was, ‘what treatment would you normally do?’ Many centres highlighted that they found answering this question difficult: most centres reported that they felt it was their duty to tell parents what their normal practice was, and said that many families preferred to have the treatment that was normally done.

Throughout the study, the participating centres were offered several different opportunities to support/improve their technique for introducing the study to families (e.g. written provision of a technique known to be successful, invitations to ask for training that they felt they needed, the opportunity to attend a customised, in-person training day). Only one centre requested further advice about introducing the study, but this was requested for the study co-ordinator rather than the PI or co-investigators. Despite acknowledging certain challenges with introducing the study to families, none of the other centres requested any training or advice, and often declined training and advice when offered – arrangements for an in-person training day were abandoned because of an overall lack of interest and uptake. Subsequently, during the individual centre visits, investigators were again asked to comment about any training needs they may have had, and their use of the successful best practice technique and study film that had been provided. Again, none of the investigators felt that any training was required, they all felt they were using the best practice and only a couple of centres actively used the video. Some centres stated that they simply were not seeing enough eligible children, thus training would have been useless. Ultimately, study recruitment would have still been well below target even if the families of the 26 eligible children who refused the study had entered; however, it does highlight the fact that there was room for improvement in how the study was being introduced to families.

The qualitative telephone interviews have offered some revealing insight into the parents’ impression of the study information process. Ethics approved, consistent information about the Hip ‘Op trial was made available to all potential participants families in two ways: (1) the patient information sheet and (2) the study information film. No feedback was given about the patient information sheet but, as reported by staff at participating centres, most parents confirmed that they had not been shown, and had not viewed, the study information film. Interestingly, the qualitative telephone interviews revealed that only those (few) participants who had viewed the study film seemed to understand the temporal focus of the study. Some interview participants went further and clearly described their feelings of confusion about the trial. It is worth noting that these responses are from families that did join the study; we have no way to assess if the families that did not participate refused because they experienced similar feelings of confusion.

Within the clinical literature there is currently no evidence clearly demonstrating which timing of surgery results in the best clinical outcome. Inevitably, individual surgeons have their own preferences for which timing is best, and parents commonly wanted to know what this preference was. Despite the lack of clear clinical evidence, responses garnered during the qualitative telephone interviews indicated that many parents had a clear preference for treatment timing that was not only based on the information and advice received, but also their own internet research, and online forums and groups where parents can discuss their experiences. Our findings here suggest that the preference of most families was for early treatment. It is likely that this preference was based on the perception of how disruptive late treatment can be to important aspects of day-to-day life (e.g. returning to work after maternity leave, finding suitable child care, day-to-day management of a larger, older child in a cast). As already stated, there is currently no clear evidence demonstrating which timing of surgery is best, neither is there any consistent or official (NHS) advice and support about the wider ‘at-home’ aspects of treatment; thus, families are in a difficult position and seem to be seeking the treatment that they perceive will be least disruptive in the short term.
The qualitative telephone interviews revealed that some parents were obviously relieved when their preferred treatment timing was allocated at randomisation, and for other parents, despite agreeing to take part in the trial, the outcome of randomisation would have affected their likelihood of remaining on the study.

During the study, two patients were withdrawn. One was withdrawn immediately after randomisation because the investigator chose to randomise them on the same day that their surgery was already planned (i.e. the investigator ‘gambled’ that the desired treatment arm would be allocated). The family of the patient may have been misinformed about the study and the randomisation was therefore invalid. This represented a serious breach of good clinical practice, the incident was fully investigated by the sponsor and the local research and development department was made aware. The other patient was withdrawn when the participant’s family sought a second opinion from a surgeon at a different centre and the surgery was performed against the timing allocated at randomisation. This patient withdrawal was also investigated by sponsor because the surgery was performed at a centre participating in the Hip ’Op trial (the same centre that incorrectly randomised the aforementioned patient), by a Hip ’Op trial investigator.

With a few notable exceptions, most centres cited the lack of suitable patients as the overwhelming reason for poor recruitment. However, several inconsistencies suggest that the reality was different: numbers provided at feasibility were much higher than actual numbers screened and no explanation was found as to why the original numbers should have been so inaccurate; some centres were screening and recruiting as expected, whereas others were not and there was no clear reason why; and many centres (particularly those that were not screening and recruiting as expected) were unenthusiastic, uncommunicative and/or defensive when efforts were made to improve recruitment and investigate why recruitment was not as expected. For these reasons, we conclude that some investigators participating in the Hip ’Op trial had difficulties with surgical equipoise and thus did not screen or recruit many, or indeed any, patients. In retrospect, it is clear that in at least one centre the lead local investigator did not have any equipoise and should not have been recruiting to the study. Surgical trials are notoriously difficult to recruit to and surgical equipoise is a major issue. The findings from the Hip ’Op trial suggest that it has suffered from the same difficulties as many other surgical trials – lack of robust and honest feasibility, lack of real commitment on the part of the PI and their team and, primarily, lack of surgical equipoise, which was of paramount importance to a study such as the Hip ’Op trial.

**Study results**

Owing to the early closure of the trial, no primary outcome data were collected, and no meaningful analysis or conclusions could be made from the very small number of secondary outcome data that were collected. In terms of safety, it is worth noting that no significant AEs occurred; this was as expected because all procedures within the trial were as per standard practice, only the timing of surgery was allocated by randomisation.

**Qualitative aspects**

This research had the potential to make a unique contribution to the evidence base for DDH interventions and to have far-reaching impacts on clinical decision-making and practice, and family support. Further funding is being sought to explore the experiences of a greater diversity of families and to examine the long-term impacts.

This pilot work has begun to highlight the wider significance of understanding parents’ experiences qualitatively. For instance, many shared their difficulties being heard, and gaining access to expert orthopaedic care, demonstrating the role of qualitative research in raising awareness of the condition among those working in primary care, particularly GPs. The outcomes are likely to aid understanding around the early diagnosis of DDH, which if identified before 8 weeks of age, increases the efficacy of non-surgical interventions. Treatment in infancy is also essential to preventing problems in later life, including osteoarthritis.
This work also has the potential to impact on secondary care practice. For example, the in-depth interview accounts detail the ways in which different parents/carers manage the process of treatment and rehabilitation. They also pinpoint broader impacts on family mobility, child care and employment, as well as implications for parental well-being. Methodologically, this pilot work, if extended further, could contribute to the growth and application of qualitative work within clinical literature, particularly paediatric orthopaedics, in which DDH is considered to be an important area of interest and qualitative research is underutilised.

Strengths and limitations

The main strength of the Hip ‘Op trial was that it was set up to answer a very important question, and it was planned and designed accordingly. Owing to early recognition of the recruitment challenges, feasibility was assessed from the point of view of both the participants and the participating centres: the response to this suggested that there was enough support from both sides and the study went ahead. The pragmatic design meant that it should have been relatively easy to conduct because all of the procedures, techniques, imaging, etc. were not dictated by the protocol – only the timing was allocated at randomisation. The key limitation of the study was that it relied on all surgeons at all centres being committed to identifying eligible children and presenting the study in a balanced manner to the children’s families. Another limitation was that many of the parents/carers of children with DDH seemed to have a clear idea of the type of treatment that they wanted for their child. Parents’ opinions about which treatment was right for their child were probably influenced by the increasing use of online information and parental DDH discussion forums, although this was not widely reported in the telephone interviews. The main limitations of the Hip ‘Op trial were beyond the control of the trial management team. The Hip ‘Op trial was set up and managed strongly: most of the sites were open in a very short space of time and a huge amount of effort was made to improve recruitment as soon as issues became apparent. Given the initial concerns about the studies ability to recruit, the 18-month pilot was written into the study design; this proved to be a strength because it established a clear cut-off point that prevented wasting resources on a failing trial. Finally, inclusion of the qualitative aspect was a great strength as it not only provided the only fruitful part of the study, but it also delivered some valuable insight into possible reasons why the trial failed.

Although the qualitative aspect achieved 14 out of the 20 (70%) participants for the planned pilot, diversity in the families who were interviewed was limited by the overall poor recruitment to the main trial. Given the multiplicity of factors in surgeon–parent interactions, aftercare, family circumstances, etc., the qualitative aspect sought to achieve data from as broad a range of participants as possible, from multiple centres, with no claims of data saturation. Had the main trial continued, the purposive sampling strategy sought to maximise the available diversity.

Lessons for the future

If we were setting up the Hip ‘Op trial again, ideally we would conduct face-to-face interviews with the team at each prospective site to investigate potential pitfalls, loopholes and concerns at the outset. Second, prior to sites opening, we would agree with the sites a standardised way in which to present the trial in an unbiased manner to minimise numbers who decline to take part. We would undertake more intensive work with patient support organisations to bring the study to the attention of parents before their first orthopaedic outpatient attendance. We would also investigate whether or not it was feasible to conduct the study in centres internationally, rather than in the UK only.

We would use the Quintet 9 (Qualitative Research Integrated within Trials) intervention, or something similar, to work with local investigators to optimise the recruitment process.
In more general terms, the exact points in which a surgical trial is susceptible to failure will vary with the clinical area, specialty and patient group. Some clinical indications will have a much richer routine data set than we had access to, so may have been able to avoid the pitfall of insufficient patients. Some specialties will have a stronger research tradition than paediatric orthopaedic surgery, have consultants with pragmatic research experience and be in a better position to access available research infrastructure. These points should inform a recruitment risk assessment for any surgical trial.

Future research

The question posed by the Hip ‘Op trial is still valid and remains unanswered; however, it is clear that it cannot currently be answered in the UK. Similar research is under way internationally, and it is likely that data from the Hip ‘Op trial could contribute to a meta-analysis from this effort. Any further UK-based trial would need to take into account the experiences of this study, and probably be planned to contribute to a meta-analysis rather than answer the primary research question on its own.

The qualitative part of the study has shed light on aspects of the DDH experience that could have far-reaching impacts on clinical decision-making and practice, and family support. Further funding is being sought to explore the experiences of a greater diversity of families and to examine the long-term impacts, as well as producing data-driven resources for families of children with DDH and clinicians, to improve support, especially during the postoperative period. On completion, the resources will be disseminated to the participants in this study.

In addition, the possibility of addressing this question using historical routinely collected data is being explored and may lead to a further project in time.
Chapter 5  Conclusion

Despite the disappointing outcome of the Hip ‘Op trial, we believe that there is a place for randomised controlled trials involving established surgical centres. Hip ‘Op has highlighted the importance of accurate advance information on numbers of available eligible patients, as well as equipoise, commitment and support from all participating investigators, when conducting surgical research and how the lack of these important elements can lead to a spectacular inability to recruit. Despite substantial consultation with parents of patients in the planning stage, the level of non-participation experienced during recruitment was much higher than anticipated. Failure of surgical trials to recruit sufficiently because of investigator equipoise issues appears to be common, and it seems that the Hip ‘Op trial has proved to be no exception. An important research question has gone unanswered, and patient care could be impacted in the long term because of this. Results of the proposed meta-analysis will prove to be very interesting and of great import to the surgical paediatric community in the future.

The Hip ‘Op trial was novel because of the inclusion of the qualitative research aspects. This part of the project has provided valuable insight into the sociological aspects of an orthopaedic intervention. The findings highlighted considerable variation in parents’ experiences in primary care, and over half of the sample experienced difficulty accessing expert orthopaedic care, resulting in multiple consultations, even when parents had a family history of DDH, before gaining onward referral.

Although families were generally positive about their child’s surgery, this study has produced detailed data about the challenges faced by families postoperatively (especially around personal care) and the techniques they developed to overcome these. DDH has significant financial implications for families, including extended parental leave, parents leaving their paid employment to care for their children, costs for hospital stays and travel, in addition to replacement equipment and furniture.

The study has underlined how important these results are, not only in terms of patient participation in clinical research, but in in terms of highlighting the (currently unmet) need for appropriate advice and robust support for parents from health-care providers regarding the ‘real-life’ aspects of managing babies and children with hip dysplasia.
Acknowledgements

The Hip ‘Op trial investigators thank the following people for their invaluable contributions:

- the Hip ‘Op trial participants and their families
- research teams at the participating centres
- the Hip ‘Op TMG, Trial Steering Committee and DMEC members (see Appendix 5 for details)
- Sue Banton, co-applicant on the Hip ‘Op trial, patient representative on the study, founder of the Steps charity
- Professor Steve Morris and Gabriella Conti (University College London) who provided the health economics expertise
- staff at the SCTU who helped along the way, particularly Marina Lee who assisted with site closedown.

The trial was run and managed by the director of Southampton Clinical Trials Unit, Professor Gareth Griffiths.

Contributions of authors

Dr Charlotte L Williams (Trial Manager at SCTU) managed the trial on a day-to-day basis for the duration of the study (from site set up to closedown) and conducted all site visits. She project managed the study closedown and report writing process. She also drafted the report and wrote many sections of it including the abstract, methods, results, discussion and conclusion (non-qualitative).

Dr Susie Weller (Research Fellow for the Qualitative Work) organised and conducted the qualitative telephone interviews, conducted the analysis for this and wrote the qualitative parts of the report. She also reviewed the draft report.

Dr Lisa Roberts (National Institute for Health Research Senior Clinical Lecturer, Associate Professor and Consultant Physiotherapist) designed the study with Nicholas Clarke, Andreas Roposch, Andrew Cook, Isabel Reading, Louise Stanton, Sue Banton, Professor Steve Morris and Wendy Wood. She designed and led the qualitative aspects of the study. She contributed to the analysis and writing of the qualitative parts of the report and reviewed report drafts.

Dr Isabel Reading (Senior Medical Statistician, Research Design Service South Central) designed the study with Nicholas Clarke, Andreas Roposch, Andrew Cook, Lisa Roberts, Louise Stanton, Sue Banton, Professor Steve Morris and Wendy Wood. She provided key statistical input during study design and conducted the final analysis of the baseline data and demographics. She also reviewed the draft report.

Dr Andrew Cook (Consultant in Public Health Medicine and Fellow in HTA) designed the study with Nicholas Clarke, Andreas Roposch, Isabel Reading, Lisa Roberts, Louise Stanton, Sue Banton, Professor Steve Morris and Wendy Wood. He drafted the report glossary and reviewed report drafts. Dr Cook co-ordinated the response to peer review.

Mrs Louisa Little (Senior Trial Manager at SCTU) was responsible for the project management of Hip ‘Op throughout the trial. She was responsible for developing and finalising the study protocol and all of the other trial documents, and working with the trial manager in the set up and initiation of the study sites. She contributed to the development of the report, drafted the Plain English summary and reviewed the draft reports.

© Queen’s Printer and Controller of HMSO 2017. This work was produced by Williams et al. under the terms of a commissioning contract issued by the Secretary of State for Health. This issue may be freely reproduced for the purposes of private research and study and extracts (or indeed, the full report) may be included in professional journals provided that suitable acknowledgement is made and the reproduction is not associated with any form of advertising. Applications for commercial reproduction should be addressed to: NIHR Journals Library, National Institute for Health Research, Evaluation, Trials and Studies Coordinating Centre, Alpha House, University of Southampton Science Park, Southampton SO16 7NS, UK.
Dr Wendy Wood (Senior Trial Manager at SCTU) designed the study with Nicholas Clarke, Andreas Roposch, Isabel Reading, Lisa Roberts, Louise Stanton, Sue Banton, Professor Steve Morris and Andrew Cook. She managed the grant application process and reviewed draft reports.

Mrs Louise Stanton (Senior Statistician at SCTU) designed the study with Nicholas Clarke, Andreas Roposch, Isabel Reading, Lisa Roberts, Wendy Wood, Sue Banton, Professor Steve Morris and Andrew Cook. She provided key statistical input during study design and oversaw the statistical elements throughout the study. She reviewed draft reports.

Professor Andreas Roposch (Consultant Orthopaedic Surgeon and Clinical Epidemiologist) designed the study with Nicholas Clarke, Andrew Cook, Isabel Reading, Louise Stanton, Sue Banton, Lisa Roberts, Professor Steve Morris and Wendy Wood. He was the co-CI and attended site visits. He reviewed the report drafts.

Professor Nicholas MP Clarke (Consultant Orthopaedic Surgeon) designed the study with Andreas Roposch, Dr Andrew Cook, Isabel Reading, Louise Stanton, Sue Banton, Lisa Roberts, Professor Steve Morris and Wendy Wood. He was the CI and the principal applicant in the effort to secure funding. He actively promoted the trial among his clinical colleagues. He reviewed the report drafts.

Data sharing statement

Data can be requested by contacting the data custodian at SCTU (ctu@soton.ac.uk). Data will be available to the scientific community with as few restrictions as feasible, following review by the Data Release Committee. Exclusive use of the data shall be retained until the publication of key outputs. The data generated from the qualitative work is not suitable for sharing beyond those contained in the report because of sensitive material content.
References


REFERENCES


## Appendix 1  Hip ’Op study centres and principal investigators

<table>
<thead>
<tr>
<th>Centre</th>
<th>Hospital trust</th>
<th>PI</th>
</tr>
</thead>
<tbody>
<tr>
<td>Alder Hey</td>
<td>Alder Hey Children’s NHS Foundation Trust</td>
<td>Colin Bruce</td>
</tr>
<tr>
<td>Barts</td>
<td>Barts Health NHS Trust</td>
<td>Manoj Ramachandran</td>
</tr>
<tr>
<td>Bristol</td>
<td>University Hospitals Bristol NHS Foundation Trust</td>
<td>Simon Thomas</td>
</tr>
<tr>
<td>Coventry &amp; Warwick</td>
<td>University Hospitals Coventry &amp; Warwickshire NHS Trust</td>
<td>Stephen Cooke</td>
</tr>
<tr>
<td>Durham</td>
<td>County Durham and Darlington NHS Foundation Trust</td>
<td>Jonathan Page</td>
</tr>
<tr>
<td>East Lancashire</td>
<td>East Lancashire Hospitals NHS Trust</td>
<td>Robin Paton</td>
</tr>
<tr>
<td>GOSH</td>
<td>Great Ormond Street Hospital for Children NHS Foundation Trust</td>
<td>Andreas Roposch</td>
</tr>
<tr>
<td>Leeds</td>
<td>Leeds Teaching Hospitals NHS Trust</td>
<td>Mohamed Sabouni</td>
</tr>
<tr>
<td>Leicester</td>
<td>University Hospitals of Leicester NHS Trust</td>
<td>Alwyn Abraham</td>
</tr>
<tr>
<td>Newcastle</td>
<td>The Newcastle upon Tyne Hospitals NHS Foundation Trust</td>
<td>Philip Henman</td>
</tr>
<tr>
<td>Nottingham</td>
<td>Nottingham University Hospitals NHS Trust</td>
<td>Kathryn Price</td>
</tr>
<tr>
<td>Oxford</td>
<td>Oxford University Hospitals NHS Trust</td>
<td>Timoleon Theologis</td>
</tr>
<tr>
<td>Plymouth</td>
<td>Plymouth Hospitals NHS Trust</td>
<td>Ben Holroyd</td>
</tr>
<tr>
<td>RNOH Stanmore</td>
<td>RNOH NHS Trust</td>
<td>Deborah Eastwood</td>
</tr>
<tr>
<td>Royal Devon and Exeter</td>
<td>Royal Devon and Exeter NHS Foundation Trust</td>
<td>Peter Cox</td>
</tr>
<tr>
<td>Sheffield</td>
<td>Sheffield Children’s NHS Foundation Trust</td>
<td>Mark Flowers</td>
</tr>
<tr>
<td>Southampton</td>
<td>University Hospital Southampton NHS Foundation Trust</td>
<td>Nicholas Clarke (CI)</td>
</tr>
<tr>
<td>Stoke-on-Trent</td>
<td>University Hospitals of North Midlands NHS Trust</td>
<td>Belen Carsi</td>
</tr>
<tr>
<td>Sunderland</td>
<td>City Hospitals Sunderland NHS Foundation Trust</td>
<td>Gavin De Kiewiet</td>
</tr>
</tbody>
</table>

GOSH, Great Ormond Street Hospital.
Appendix 2  Topic guide for telephone interview with parents

Questions for semi structured telephone interview with people with parents, with probes or follow up questions.

Topics to explore first with parents  [Background/setting the scene]

Before we talk about what it was like for you taking part in the trial, I would like to ask you some background questions about how your child’s hip condition impacts upon your day to day life.

• So, firstly, have you had any previous experience of a child with a hip condition?  [Probes: other siblings, self, others]

• Could you describe for me what effect ____ (name) ____’s hip condition had on him/her before the surgery?

• Could you describe for me how you think ____ (name) ____’s hip condition impacts on your day to day, family life?  [Probes: mood, sleep, mobility, play, relationship with you, siblings, others?]

• What, if any, personal costs do you think you have experienced as a result of ____ (name) ____ hip condition?  [Probes: financial e.g. time off work, special equipment]

• What were your expectations of the surgery?

Topics to explore about the surgery

Before we talk about how things are now, I would like to ask you about your experience of ____ (name)’s ____ hip surgery:

• What went well for you?

• What could have been improved?

• Is there anything you know now that you wish you’d known before the surgery?

• How have things been since the surgery?

• Have you noticed any changes [related to the topics outlined above] since the surgery?

• Has there been any change in family life since the surgery?  [Probes: topics covered earlier]

• Have you had any additional personal costs to cover following the surgery?  [Probes: time off work, equipment etc]

• Do you have any advice for the surgery team from your point of view?

• If a friend or family member was in the same position with a child experiencing this hip condition, what advice would you give them?

• Have your expectations about the surgery changed?
Looking forward

- Thinking about the future, what do you think the outcome is going to be for _____(name)_____?

- Do you have any worries or concerns about _____(name)______ for the future? [probe: short-term, medium-term and long-term]

Experience of the trial

- Firstly, could you tell me what made you decide to take part in the trial?

- Did you have any worries or concerns before it started? If so, what was it that concerned you?

- What was it like to take part? [probe: experience of questionnaire, surgery, interview]

- So, thinking about the overall experience was it as you expected it would be?

- Do you have any advice for the research team from your point of view?

- Is there anything that we haven’t talked about that you would like to mention?
Appendix 3 Observations, actions and outcomes in relation to the management of the recruitment issues encountered during the Hip ‘Op trial throughout 2015

<table>
<thead>
<tr>
<th>Month (2015)</th>
<th>Observation</th>
<th>Action</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>January</td>
<td>Recruitment on track</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>February</td>
<td>Recruitment slightly below target</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>March</td>
<td>Recruitment issue identified:</td>
<td>TMG discussion:</td>
<td>No effect on recruitment</td>
</tr>
<tr>
<td></td>
<td>• fewer than expected entries on screening logs</td>
<td>• CI correspondence with all site PIs</td>
<td></td>
</tr>
<tr>
<td></td>
<td>• eligible patients refusing trial</td>
<td>• Decision SCTU should:</td>
<td></td>
</tr>
<tr>
<td></td>
<td>HTA programme informed</td>
<td>• contact sites – highlight importance of appropriate introduction of study to families</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>• organise TC with site teams to discuss screening</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>• Steps – reminder to highlight/promote study on website/newsletters</td>
<td></td>
</tr>
<tr>
<td>April</td>
<td>Recruitment below target</td>
<td>• SCTU – push to open remaining sites</td>
<td>No effect on recruitment</td>
</tr>
<tr>
<td></td>
<td></td>
<td>• Reminder to use DVD to support patient information process</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>• Encouraged to contact SCTU for support with recruitment issues</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>• SCTU suspected that sites had overestimated patient population in original application</td>
<td></td>
</tr>
<tr>
<td>May</td>
<td>Recruitment below target</td>
<td>• SCTU – push to open remaining sites</td>
<td>Nine sites reply to survey e-mail; no effect on recruitment</td>
</tr>
<tr>
<td></td>
<td></td>
<td>• Recruitment survey e-mail to sites – request for detailed answers focusing on the questions:</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>๏ Why are so few patients being screened?</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>๏ What is the most successful way to introduce the study to families?</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>• SCTU informed sites as part of survey e-mail that a meeting was being planned with sites to discuss recruitment</td>
<td></td>
</tr>
<tr>
<td>June</td>
<td>Recruitment below target</td>
<td>• SCTU – push to open remaining sites</td>
<td>No effect on recruitment; positive response from TC participants but no effect on recruitment</td>
</tr>
<tr>
<td></td>
<td></td>
<td>• TMG meeting: recruitment recovery main focus of discussion</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>๏ Nicholas Clarke’s successful method of recruitment sent to all sites (sent 4 June 2015)</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>๏ TC with RNs from all sites and PPI TMG member (held 10 June 2015)</td>
<td></td>
</tr>
<tr>
<td>Month (2015)</td>
<td>Observation</td>
<td>Action</td>
<td>Outcome</td>
</tr>
<tr>
<td>-------------</td>
<td>-------------</td>
<td>--------</td>
<td>---------</td>
</tr>
<tr>
<td></td>
<td></td>
<td>• SCTU:</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>○ began arrangements for training meeting – poor uptake by site teams</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>○ investigated possibility of Q-QAT involvement</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>○ created study-specific website (went live 26 September 2015)</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>○ investigated producing site staff training video</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>○ provided desktop reminder cards to all sites – DMEC recommendation (sent 29 June 2015)</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>○ reminded all sites that individual support and training could be provided by SCTU/Southampton RNs</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>• HTA programme informed of the above</td>
<td></td>
</tr>
<tr>
<td>July</td>
<td>Recruitment below target</td>
<td>• SCTU – push to open remaining sites</td>
<td>–</td>
</tr>
<tr>
<td></td>
<td></td>
<td>• SCTU:</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>○ introduced pre-screening logs</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>○ began preparations for the training meeting – meeting content (agenda, materials, etc.) and meeting date (contact with all sites, diaries, etc.)</td>
<td></td>
</tr>
<tr>
<td>August</td>
<td>Recruitment below target</td>
<td>• Provisional arrangements for training meeting abandoned because of:</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>○ poor uptake by site teams</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>○ strengthened awareness that recruitment issues seem to stem from a lack of suitable patients</td>
<td></td>
</tr>
<tr>
<td>September</td>
<td>Recruitment below target</td>
<td>• SCTU – push to open remaining sites</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>• The Hip ‘Op trial was promoted by TSC PPI member on her public blog and in the press release for her book launch</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>• Website went live</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>• Commenced arrangements for individual site visits</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>• Q-QAT informed that services not required</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>No effect on recruitment</td>
<td></td>
</tr>
<tr>
<td>October</td>
<td>Recruitment below target</td>
<td>• SCTU – push to open remaining sites</td>
<td>–</td>
</tr>
<tr>
<td></td>
<td></td>
<td>• Site visits by co-CI and trial manager</td>
<td></td>
</tr>
<tr>
<td>November</td>
<td>Recruitment below target</td>
<td>• SCTU – push to open remaining sites</td>
<td>–</td>
</tr>
<tr>
<td></td>
<td></td>
<td>• Site visits by co-CI and trial manager</td>
<td></td>
</tr>
<tr>
<td>December</td>
<td>Recruitment below target</td>
<td>• SCTU – push to open remaining sites</td>
<td>–</td>
</tr>
<tr>
<td></td>
<td></td>
<td>• Site visits by co-CI and trial manager</td>
<td></td>
</tr>
</tbody>
</table>

DVD, digital versatile disc; PPI, patient and public involvement; RN, research nurse; TC, teleconference; TSC, Trial Steering Committee.
### Appendix 4  Details of participants who were withdrawn and lost to follow-up

<table>
<thead>
<tr>
<th>Allocation</th>
<th>Withdrawn/lost to follow-up</th>
<th>Reason</th>
<th>Received surgery?</th>
</tr>
</thead>
<tbody>
<tr>
<td>Delayed</td>
<td>Lost to follow-up</td>
<td>The participant’s family did not return to clinic</td>
<td>Unknown</td>
</tr>
<tr>
<td>Delayed</td>
<td>Withdrawn</td>
<td>The participant was the subject of a serious breach. Randomisation was invalid</td>
<td>Yes (but not according to allocated timing)</td>
</tr>
<tr>
<td>Delayed</td>
<td>Withdrawn</td>
<td>The patient’s family sought another opinion and went ahead with surgery – the PI where the patient was randomised was not aware</td>
<td>Yes (but in a different centre not according to allocated timing)</td>
</tr>
<tr>
<td>Early</td>
<td>Lost to follow-up</td>
<td>Patient was lost to follow-up at 9 months post surgery – reason unknown</td>
<td>Yes</td>
</tr>
</tbody>
</table>
Appendix 5  Governance committee membership

Trial Steering Committee, Data Monitoring and Ethics Committee and Trial Management Group members

**Trial Steering Committee**

- Mr Michael Bell (chairperson).
- Professor James Kasser.
- Professor Jan Van Der Meulen.
- Ms Ly-Mee Yu.
- Ms Natalie Trice (patient and public representative).

**Data Monitoring and Ethics Committee**

- Mr Michael Benson (chairperson).
- Miss Ines Rombach.
- Mr Pablo Castañeda.

**Trial Management Group**

- Professor Nicholas Clarke (orthopaedic surgeon and CI).
- Professor Andreas Roposch (orthopaedic surgeon and co-CI).
- Ms Belen Carsi (orthopaedic surgeon and PI).
- Mr Colin Bruce (orthopaedic surgeon and PI).
- Mrs Louisa Little (SCTU senior trials manager).
- Mr Ian Ratcliffe (SCTU statistician).
- Dr Lisa Roberts (qualitative researcher).
- Mrs Sue Banton (patient and public representative).
- Dr Charlotte Williams (SCTU trials manager).
- Mrs Liz Blake (research nurse representative).
Appendix 6  Patient and public involvement in the Hip ‘Op trial

Sue Banton, a patient support group representative, was actively involved in the development of the Hip ‘Op trial. Her knowledge and expertise was used in the planning and design stages of the project and continued throughout the ‘life’ of the trial. She was involved in the development of the trial documents, including the patient information sheet and the information digital versatile disc (DVD). She was a co-applicant on the trial grant and an active member of the TMG. She attended as many meetings as possible, where she shared her knowledge and expertise in the management of DDH and the associated impact that treatment for DDH has on the families of the children affected. Her input into the qualitative aspects of the trial was invaluable to the qualitative researchers.

Sue was a founder of Steps, a national charity supporting children and adults affected by lower limb conditions. In the developmental stages of the project, through Steps, Sue conducted an ‘acceptability’ questionnaire using a social media site, identifying how best to help parents to decide whether or not to participate in research. The outcome included accessible information for wider family members and the importance of a clear explanation of the trial from a senior health-care professional. At the outset, it was planned that Steps would have a wider part to play in the trial by offering support and disseminating the findings of the trial to families via their website, newsletters, social media and meetings; however, as a result of internal issues, Steps were unable to fully engage as planned.

A parent/carer of a child with DDH, Kirsten Armstrong was fundamental in assisting with the development of the patient information DVD, and it was originally intended that she would continue to be involved in the trial and join the review group and the TMG. However, she was unable to commit to this and Sue Banton continued instead in the role of TMG member as the dedicated patient and public involvement (PPI) representative throughout the trial.

Natalie Trice joined the Trial Steering Committee as PPI representative 8 months after the Hip ‘Op trial opened to recruitment. Natalie is the mother of a child with DDH. In addition, she is a DDH blogger and recently published a book about her experiences. She helped raise awareness of the Hip ‘Op trial on her blog and at the launch of her book.
## Appendix 7 The consolidated criteria for reporting qualitative studies: 32-item checklist

<table>
<thead>
<tr>
<th>Number of item</th>
<th>Guide questions/description</th>
<th>Reported on page number</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Domain 1: research team and reflexivity</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>Personal characteristics</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>1. Interviewer/facilitator</td>
<td>Which author/s conducted the interview or focus group?</td>
<td>65</td>
</tr>
<tr>
<td>2. Credentials</td>
<td>What were the researcher’s credentials (e.g. PhD, MD)?</td>
<td>65</td>
</tr>
<tr>
<td>3. Occupation</td>
<td>What was their occupation at the time of the study?</td>
<td>65</td>
</tr>
<tr>
<td>4. Gender</td>
<td>Was the researcher male or female?</td>
<td>24, 65</td>
</tr>
<tr>
<td>5. Experience and training</td>
<td>What experience or training did the researcher have?</td>
<td>24</td>
</tr>
<tr>
<td><strong>Relationship with participants</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>6. Relationship established</td>
<td>Was a relationship established prior to study commencement?</td>
<td>9</td>
</tr>
<tr>
<td>7. Participant knowledge of the interviewer</td>
<td>What did the participants know about the researcher (e.g. personal goals, reasons for doing the research)?</td>
<td>9</td>
</tr>
<tr>
<td>8. Interviewer characteristics</td>
<td>What characteristics were reported about the interviewer/facilitator (e.g. bias, assumptions, reasons and interests in the research topic)?</td>
<td>Field notes, etc., p. 9</td>
</tr>
<tr>
<td><strong>Domain 2: study design</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>Theoretical framework</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>9. Methodological orientation and theory</td>
<td>What methodological orientation was stated to underpin the study (e.g. grounded theory, discourse analysis, ethnography, phenomenology, content analysis)?</td>
<td>Thematic analysis/framework, p. 9</td>
</tr>
<tr>
<td><strong>Participant selection</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>10. Sampling</td>
<td>How were participants selected (e.g. purposive, convenience, consecutive, snowball)?</td>
<td>9</td>
</tr>
<tr>
<td>11. Method of approach</td>
<td>How were participants approached (e.g. face to face, telephone, mail, e-mail)?</td>
<td>9</td>
</tr>
<tr>
<td>12. Sample size</td>
<td>How many participants were in the study?</td>
<td>24</td>
</tr>
<tr>
<td>13. Non-participation</td>
<td>How many people refused to participate or dropped out? Reasons?</td>
<td>Table 8, p. 24</td>
</tr>
<tr>
<td><strong>Setting</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>14. Setting of data collection</td>
<td>Where was the data collected (e.g. home, clinic, workplace)?</td>
<td>By telephone, p. 9</td>
</tr>
<tr>
<td>15. Presence of non-participants</td>
<td>Was anyone else present besides the participants and researchers?</td>
<td>rVa</td>
</tr>
<tr>
<td>16. Description of sample</td>
<td>What are the important characteristics of the sample (e.g. demographic data, date)?</td>
<td>25</td>
</tr>
<tr>
<td><strong>Data collection</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>17. Interview guide</td>
<td>Were questions, prompts, guides provided by the authors? Was it pilot tested?</td>
<td>9 and Appendix 2</td>
</tr>
<tr>
<td>18. Repeat interviews</td>
<td>Were repeat interviews carried out? If yes, how many?</td>
<td>rVa</td>
</tr>
<tr>
<td>Number of item</td>
<td>Guide questions/description</td>
<td>Reported on page number</td>
</tr>
<tr>
<td>----------------</td>
<td>-------------------------------------------------------------------------------------------</td>
<td>-------------------------</td>
</tr>
<tr>
<td>19.</td>
<td>Audio/visual recording Did the research use audio or visual recording to collect the data?</td>
<td>9</td>
</tr>
<tr>
<td>20.</td>
<td>Field notes Were field notes made during and/or after the interview or focus group?</td>
<td>9</td>
</tr>
<tr>
<td>21.</td>
<td>Duration What was the duration of the interviews or focus group?</td>
<td>Table 9, p. 25</td>
</tr>
<tr>
<td>22.</td>
<td>Data saturation Was data saturation discussed?</td>
<td>61</td>
</tr>
<tr>
<td>23.</td>
<td>Transcripts returned Were transcripts returned to participants for comment and/or correction?</td>
<td>n/a</td>
</tr>
</tbody>
</table>

**Domain 3: analysis and findings**

**Data analysis**

<table>
<thead>
<tr>
<th>Number of item</th>
<th>Guide questions/description</th>
<th>Reported on page number</th>
</tr>
</thead>
<tbody>
<tr>
<td>24.</td>
<td>Number of data coders How many data coders coded the data?</td>
<td>24</td>
</tr>
<tr>
<td>25.</td>
<td>Description of the coding tree Did authors provide a description of the coding tree?</td>
<td>Framework, p. 9</td>
</tr>
<tr>
<td>26.</td>
<td>Derivation of themes Were themes identified in advance or derived from the data?</td>
<td>24</td>
</tr>
<tr>
<td>27.</td>
<td>Software What software, if applicable, was used to manage the data?</td>
<td>9</td>
</tr>
<tr>
<td>28.</td>
<td>Participant checking Did participants provide feedback on the findings?</td>
<td>12</td>
</tr>
</tbody>
</table>

**Reporting**

<table>
<thead>
<tr>
<th>Number of item</th>
<th>Guide questions/description</th>
<th>Reported on page number</th>
</tr>
</thead>
<tbody>
<tr>
<td>29.</td>
<td>Quotations presented Were participant quotations presented to illustrate the themes/findings? Was each quotation identified (e.g. participant number)?</td>
<td>25–56</td>
</tr>
<tr>
<td>30.</td>
<td>Data and findings consistent Was there consistency between the data presented and the findings?</td>
<td>25–56</td>
</tr>
<tr>
<td>31.</td>
<td>Clarity of major themes Were major themes clearly presented in the findings?</td>
<td>24</td>
</tr>
<tr>
<td>32.</td>
<td>Clarity of minor themes Is there a description of diverse cases or discussion of minor themes?</td>
<td>25–26</td>
</tr>
</tbody>
</table>

MD, doctor of medicine; n/a, not applicable; PhD, doctor of philosophy.
Appendix 8  Changes to protocol

The Hip 'Op protocol underwent several changes during the course of the project. All were approved by the appropriate governance committees, ethics committee and funder. They are summarised in Table 10.

TABLE 10  Changes to protocol

<table>
<thead>
<tr>
<th>Version</th>
<th>Date</th>
<th>Changes from previous version</th>
</tr>
</thead>
<tbody>
<tr>
<td>V1</td>
<td>12 May 2014</td>
<td>Original protocol</td>
</tr>
</tbody>
</table>
| V2      | 28 November 2014 | 1. Amendment of the schedule of events table and clarification in the text when ultrasound or X-ray should take place, and removed CT and MRI as it was felt this was not required  
2. A secondary objective was added: ‘Presence or absence of ossific nucleus at time of primary treatment for dysplasia’  
3. Health economics and qualitative analysis was added to the secondary end points of the study, as we noted that these had been omitted in V1  
4. It was noted that the words ‘prevalence’ and ‘incidence’ had been used interchangeably throughout the protocol. The study statisticians advised we change all to ‘incidence’  
5. Terminology for ‘pre-existing conditions’ has been changed as this was also causing confusion. The terminology has been changed to make it clear that only ‘significant’ existing medical conditions should be reported  
6. Typographic errors were corrected and textual clarification of some statements have been made  
7. Appendix 3, Classification of AVN – further explanation of each group within the classification has been given and graphic imaging added |
| V3      | 13 May 2015   | 1. Trial synopsis (rationale and inclusion criteria, p. 8) and section 4.3 (Inclusion criteria, p. 15) – minimum age for inclusion stated as 12 weeks rather than 3 months  
2. Trial synopsis (inclusion criteria, p. 8) and section 4.3 (Inclusion criteria, p. 15) – the following inclusion criterion has been added: ‘children born at ≥30-weeks gestation can be included’  
3. Trial synopsis (exclusion criteria, p. 8) and section 4.3 (exclusion criteria, p. 16) – the following exclusion criteria has been added: ‘Children born at <30-weeks gestation’  
4. Section 4.5 (randomisation procedure, p. 16). The following statement has been added: ‘Eligible premature babies should not be randomised until they reach 12 weeks of age as calculated using their corrected date of birth. However, the actual date of birth of such children should be entered into the TENALEA system for randomisation’  
5. Section 5.2 (treatment regimens, p. 17). The following statement has been added: ‘or other definitive imaging’. The statement, ‘CT scan (or MRI)’ has been replaced by this statement, ‘imaging’  
6. Section 2 (trial objectives, health Technologies being assessed, p. 14) – the following statement has been removed: ‘The randomisation will allocate whether the surgical reduction is carried out immediately (within 2 weeks) or delayed until after the appearance of the ossific nucleus’  
7. Section 5.2 (treatment regimens, arm A – early treatment, p. 17) – the following statement has been added: ‘Timeframe for performing surgery is as per standard practice’  
8. Section 5.2 (treatment regimens, arm B – delayed treatment, p. 17) – the following statement has been added: ‘Surgery should take place within 2–4 weeks of the appearance of the ON (unless exceptional circumstances require it to be delayed further)’  
9. Section 5.2 (treatment regimens, arm B – delayed treatment, p. 17) – the bracketed statement ‘(not beyond 13 months)’ has been removed  
10. Section 7.1 (data collection, p. 20) – the following statement has been added: ‘where assessable from the type of imaging performed’  
11. Schedule of observations and events (table, p. 11). The points from the pre-surgery column have been moved to the clinic visit/consent column for the early cost diary, CarerQol and PedsQL. Superscript point 4 has been added for clarification  
12. Correction of various minor typographic and grammatical errors, and minor clarifications throughout |
### TABLE 10 Changes to protocol (continued)

<table>
<thead>
<tr>
<th>Version</th>
<th>Date</th>
<th>Changes from previous version</th>
</tr>
</thead>
<tbody>
<tr>
<td>V4</td>
<td>22 October 2015</td>
<td>1. Trial synopsis (primary trial endpoints, p. 7) and section 3.1 (trial endpoints, p. 13). AVN will be classified radiologically according to the Kalamchi and MacEwen grading as part of routine assessment (grade I to IV), rather than grades II to IV</td>
</tr>
<tr>
<td></td>
<td></td>
<td>2. Section 3.0 (trial design, p. 13). The following statement has been changed from, ‘A total of 636 children aged 3–13 months . . .’, to read, ‘A total of 636 children aged 12 weeks–13 months . . .’</td>
</tr>
<tr>
<td></td>
<td></td>
<td>3. Section 5.2 (treatment regimens, p. 13). The following statement has been reworded from, ‘6-weekly ultrasound (or X-ray – as per local practice) . . .’ to, ‘Ideally, 6-weekly imaging (e.g. ultrasound/X-ray – as per local practice) . . .’</td>
</tr>
<tr>
<td></td>
<td></td>
<td>4. Correction of various minor typographic and grammatical errors throughout</td>
</tr>
</tbody>
</table>

CT, computerised tomography; MRI, magnetic resonance imaging.