Development of a core outcome set for lower limb orthopaedic surgical interventions in ambulant children and young people with cerebral palsy

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List of Abbreviations

Abbreviations	Full name
10-MWT	Ten-Minute Walk Test
3DGA	Three-Dimensional Gait Analysis
ADL	Activities Of Daily Living
AEs	Adverse Events
CASP	Critical Appraisal Skills Programme
CAT	Computerised Adaptive Tests
CHQ	Child Health Questionnaire
COMET	Core Outcome Measures in Effectiveness Trials
COS	Core Outcome Set
COS-STAD	Core Outcome Set-STAndards for Development
COS-STAR	Core Outcomes Set-STAndards for Reporting
COS-STAP	Core Outcome Set-STAndardised Protocol
COSMIN	COnsensus-based Standard for the selection of health status Measurement Instruments
CP	Cerebral Palsy
ENTREQ	Enhancing Transparency in Reporting the Synthesis of Qualitative Research
EVGS	Edinburgh Visual Gait Scale
FIM	Functional Independence Measure for Children
FMS	Functional Mobility Scale
GDI	Gait Deviation Index
GGI	Gillette Gait Index
Gillette FAQ	Gillette Functional Assessment Questionnaire
GMFCS	Gross Motor Function Classification System
GMFM	Gross Motor Function Measure
GMPM	Gross Motor Performance Measure
GOAL	Gait Outcomes Assessment List
GPS	Gait Profile Score
ICF	International Classification of Functioning
ICF-CY	International Classification of Functioning- Children and Youth
IGA	Instrumented Gait Analysis
LAD	Lever Arm Dysfunction
MobQues47	Mobility Questionnaire 47
OGA	Observational Gait Analysis

OGS	Observational Gait Scale
OMERACT	Organisers of the Outcome Measures in Rheumatology
PEDI	Pediatric Evaluation of Disability Inventory
PedsQL	Paediatric QoL Inventory
PODCI	Pediatric Outcomes Data Collection Instrument
PPI	Patient and Public Involvement
PRISMA	Preferred Reporting Items for Systematic reviews and Meta- Analyses
PRISMA-ScR	Preferred Reporting Items for Systematic reviews and Meta- Analyses- Scoping Review
PROMIS	Patient-Reported Outcomes Measurement Information System
PROMs	Patient-Reported Outcome Measures
PROs	Patient-Reported Outcomes
PRS	Physician Rating Scale
QES	Qualitative Evidence Synthesis
QoL	Quality of Life
SEMLS	Single-Event Multi-Level Surgery
TUG	Timed Up and Go
VGAS	Visual Gait Assessment Scale
VGS	Visual Gait Score
WHO	World Health Organisation

Abstract

Background: The literature on lower limb orthopaedic surgery for ambulant children with cerebral palsy (CP) demonstrates significantly heterogenous outcomes and outcome measures and evidence of selective outcome reporting. This limits evidence synthesis and undermines research findings through outcome reporting bias. Although several outcomes are commonly measured and reported, these do not always capture the needs and expectations of children and families.

Purpose: The aims of this project were to (1) develop a minimum set of important and relevant outcomes, a "Core Outcome Set" for use in future clinical studies evaluating lower limb orthopaedic surgery for ambulant children with CP, and (2) develop a recommendation of optimal outcome measures, a "Core Outcome Measurement Set", to assess each outcome within the core outcome set.

Methods: The project was designed in four parts. (1) potential outcomes were identified through systematic reviews; (2) interviews with stakeholders (healthcare professionals, researchers, children and families) were undertaken; (3) an international two-round e-Delphi consensus survey was used to gain agreement between stakeholders as to which outcomes were "core"; (4) outcomes selected through the Delphi process were brought forward to a consensus meeting to agree on the final core outcome set. Target stakeholders from relevant national and international networks were approached to contribute to this selection.

The selection of core outcome measurement instruments was designed in four parts. (1) conceptual considerations; (2) identification of existing outcome measures; (3) quality assessment; and (4) development of generic recommendations on the selection of outcome measurement instruments for a COS.

Results: The literature review and stakeholder interviews initially identified 99 outcome domains. These were grouped into 41 outcomes for prioritisation in the Delphi survey. A total of 197 participants from 13 countries were surveyed. The findings were brought forward to the consensus meetings with 21 international stakeholders. Consensus was achieved on eight outcome domains: (1) pain and fatigue; (2) lower limb structure; (3) motor function; (4) daily life activities; (5) gait-related outcomes; (6) physical activity; (7) independence; and (8) quality of life. An adverse event domain was agreed as an essential part of any study documentation.

The outcome measures recommended by the consensus group were a combination of clinician-driven objective measures: (1) Three-Dimensional Gait Analysis (3-DGA); (2) Edinburgh Visual Gait Score (EVGS); (3) 10-Meter Walk Test (10-MWT); and (4) Gross Motor Functional Measure (GMFM-66), and patient-reported outcome measures (PROMs): (5) Gait Outcomes Assessment List (GOAL); (6) Gillette Functional Assessment Questionnaire (Gillette FAQ); (7) Patient-Reported Outcomes Instruments System (PROMIS); and (8) Cerebral Palsy - Quality of Life (CP-QoL).

Conclusions: This project has developed an evidence-based, internationally endorsed core outcome set for lower limb orthopaedic surgery in ambulant children with CP. It is recommended that these core sets be implemented into future clinical studies. The use of these core sets would enhance the consistency and quality of studies in orthopaedic surgery for children with CP.

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Chapter 1 Introduction

Cerebral Palsy (CP) is considered to be the most common cause of childhood disability; its global prevalence is 2-2.5 children per 1,000 (1). CP is a permanent movement and posture disorder that limits activity and is often accompanied by other non-motor disturbances (e.g., sensation, perception, cognition, communication, behaviour and seizure disorders) as well as secondary musculoskeletal problems (2). The clinical presentation varies from mild to severe impairment in motor function, which leads to mobility levels that vary from ambulatory to non-ambulatory; two thirds of children with CP are ambulatory (3).

Children with CP develop musculoskeletal deformities as a result of impaired motor control, further compromising motor function. Lower limb orthopaedic surgery is considered a standard treatment for ambulatory children (3, 4). However, the evidence to support the effectiveness of paediatric orthopaedic surgery in CP is poor quality because studies around CP involve several challenges (5-7). Firstly, the condition is heterogeneous in aetiology and manifestations, so research into relevant therapies is difficult because the treatment effects are different for each child and generalisability of the results is limited. Secondly, the variability in outcomes and outcome measures used in published studies makes comparing results between studies difficult, making systematic reviews and/or meta-analyses difficult to conduct (5, 6). Third, the relevance of the outcomes used is often not in line with what the children or families need. There is evidence that researchers, clinicians, families and patients have different perspectives regarding different treatments (7). Fourth, reviews have identified selective outcome reporting bias in the literature regarding lower limb orthopaedic surgery in CP, with adverse events reported only in a limited number of studies. This may limit the possibility for rigorous assessment of an intervention (5, 6). Finally, the published non-randomised studies in the field of CP and Single-Event Multiple Level Surgery (SEMLS), which is defined as the combination of a minimum of two operations undertaken bilaterally at a minimum of two levels (e.g., hip and knee), are of poor quality, with most studies classified as an OXFORD level III (cohort follow-up study) and only one study classified as an OXFORD level II (observational study) (5). Although researchers are moving toward randomised controlled trials (RCTs) in paediatric orthopaedic research, Dulai et al. (8) found that only 19% of published RCTs meet the criteria of high-quality studies.

The primary motivation for this research project was to explain and resolve these challenges. The efficiency, relevance, transparency and overall quality of research into the orthopaedic treatment of CP

could be increased through the development of a core outcome set (COS). This COS would form an agreed set of the most important outcomes and outcome measures that should be used and reported in all clinical trials (9).

To date, no COS has been developed for lower limb orthopaedic surgery performed on ambulant children and young people with CP. To fill this gap, I led the development of a COS for lower limb orthopaedic surgery for children and young people with CP, and I describe this process and the outcome in detail in this thesis. I adopted the evidence- based methodology that is endorsed by the Core Outcome Measures in Effectiveness Trials (COMET) and COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) initiatives (10, 11).

In this DPhil thesis, the core set describes the important outcome domains and measures for lower limb orthopaedic surgery for ambulant children and young people with CP, their caregivers and health professionals. The focus of this project is on ambulatory children with CP. Approximately two-thirds of children with CP are ambulant (1). Even in ambulatory children and adolescents with CP, walking ability is usually affected. As a result, research efforts have been directed toward improving or maintaining the young persons' walking ability through orthopaedic surgery or other interventions.

The core set of outcomes of importance will be based on the World Health Organisation (WHO) International Classification of Functioning - Children and Youth (ICF-CY) Framework (12). The ICF-CY offers a framework to facilitate comprehensive understanding of function and disability from a bio-psychosocial perspective. This framework comprises four components: 1) body functions and body structures; 2) activities and participation; 3) environmental factors and 4) personal factors.

The overall objective of the work described in this thesis was to identify which ICF-CY categories best represented the important outcomes that were sought post orthopaedic surgery of the lower limbs in ambulant children and young people with CP, aged between 8 to 18 years old at the time of the surgery. Specifically, the evidence from the perspectives of health professionals who were involved with these children and young people (i.e. researchers, clinicians), the children and young people themselves and those who cared for a child with ambulant CP were gathered, to identify which outcomes were considered the most relevant and important to measure after this treatment. Subsequently, this evidence was presented to an international group of experts and children and family representatives,

who relied on their expertise to select the final outcome domains and outcome measures to be included in the COS.

Research project question

The work that was undertaken for this DPhil project was designed to answer the following two questions:

1. What are the key health areas (outcome domains) that are relevant to health professionals, the ambulant children and young people and their caregivers when lower limb orthopaedic surgery for CP is considered?
2. What are the best outcome measures (tools) available to assess identified outcomes after surgery?

Introduction to this DPhil thesis

The thesis is presented in 12 chapters. Chapter 1 is this general introduction. Chapter 2 and Chapter 3 contain the literature review that was conducted to define CP and explore the current status of lower limb orthopaedic surgery that is performed to improve its effects. Chapter 4 presents the overall aims, objectives and describes the rationale for this DPhil project. Chapter 5 explains the study design to develop the COS*; Chapter 6 describes the qualitative evidence synthesis that focuses on the children's and parents' perspectives of the outcomes that are sought following surgery*. Chapter 7 contains a scoping review of outcome domains that have been reported in the literature on lower limb orthopaedic surgery for ambulant children with CP*. Chapter 8 presents qualitative research undertaken by conducting individual interviews with a stakeholders' group*. The interviews focused on identification of key health measures (outcome domains) that these stakeholders considered to be important goals of the surgery. Furthermore, this chapter focused on identification of key facilitators and barriers that may impact these expectations. Chapter 9 and Chapter 10 explain the consensus procedure that was undertaken to reach an agreement on the outcome domains to be included in the COS*. Chapter 11 describes the consensus procedure that was followed to agree which outcome measures should be included in the core measurements set. Chapter 12 contains the general discussion of the findings of the work described in this thesis and how results of clinical trials in CP surgery should be interpreted using the COS. It also contains suggestions for future research on lower limb surgery in children and young people with CP. Additionally, Chapter 12 presents a final conclusion.

* Chapter was adopted and extracted from published papers.

Chapter 2 Cerebral Palsy

This chapter describes a thorough review of the literature relevant to the main subjects covered in this thesis. It includes the definition of CP, the functional assessment of children with CP and the lower limb surgical treatment undertaken for this population.

2.1 Cerebral Palsy

This section provides an overview of CP, including definition, prevalence, risk factors, aetiology and differential diagnosis. The available classification systems and clinical manifestations are described together with the natural history of CP, in terms of motor function and gait. Finally, this section presents the clinical assessment of CP and the role of orthopaedic surgery in the treatment of musculoskeletal pathology of the lower limbs.

2.1.1 Definition

CP is a neurodevelopmental condition that begins in early childhood and persists through the person's lifespan (2, 4). It has always been a challenge to define 'cerebral palsy'; this is shown by the number of attempts that have been made over the years (2). In response to the heterogeneity of disorders and the limitation of the descriptions that could be covered by the previous definitions, Rosenbaum et al. (2) in 2006, slightly modified the definition to reflect the concept that CP is one group of neurodevelopmental disorders that affect numerous developing functions. The definition became: **"Cerebral Palsy describes a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of CP are often accompanied by disturbances of sensation, perception, cognition, communication, and behaviour, by epilepsy, and by secondary musculoskeletal problems"** (2).

2.1.2 Prevalence

CP is the most common cause of physical disability in children (1). CP prevalence is remarkably similar across developed countries, with an overall prevalence is 1.5 to 2.5 per 1,000 live births (13). Three reviews on the prevalence of CP have been published recently (14-16). The first, published in 2007, looked at both the overall prevalence of CP and the prevalence reported by birth weight in pre-term infants. The overall estimate of median prevalence that was reported was 2.4 per 1000 live births. In this review, only a single bibliographic database (PubMed) was searched (14).

A second systematic review, published in 2008, examined the prevalence of CP in relation to gestational age only. It demonstrated a significant decrease in the prevalence of CP with increasing gestational age. The prevalence was 146 (95% confidence interval [CI] 125-170) per 1000 children who were born at 22 to 27 weeks of gestation and steadily declined thereafter to an estimated 62 (95% CI 49-78) per 1000 children born at 28 to 31 weeks, 7.0 (95% CI 6-9) per 1000 at 32 to 36 weeks' gestation, and 1.13 (95% CI 0.93-1.14) per 1000 in term-born infants. This systematic review was carried out with rigorous methodology; four bibliographic databases were searched (MEDLINE, CINAHL, Cochrane library, and Science Direct) with a clearly reported search strategy (15).

In another review, Oskoui et al. (16) in 2013 considered the prevalence of CP among children born in 1985 or after (16). They searched two databases (MEDLINE and EMBASE) for studies published between 1985 and 2011. The review showed that the pooled overall prevalence of CP was 2.11 per 1000 live births (95% CI 1.98-2.25).

A UK study, which used a population-based register, showed that the overall CP rate among neonatal survivors fell from 2.5 per 1000 between 1984 and 1986 to 1.7 between 1993 and 1995 (17). Another study of five active CP registers in the UK showed that the mean annual prevalence rate remained at approximately 2 per 1000 live births during the birth years 1986 to 1996, with 91% of these cases recorded as spastic CP (18). Moreover, a recent study showed that the estimated number of children aged three to 15 years who were living with CP in England and Wales was about 20,500 in 2013 rising to approximately 22,100 by 2020, a 7.5 % increase (1).

2.1.3 Risk Factors and Aetiology

CP is caused by factors affecting the Central Nervous System (CNS) in the early stages of development: during pregnancy, at birth or after birth (19). Aetiological research concentrates mainly on the

identification of the pre-, peri- and post-natal risk factors, the combination of which could lead to CP (20-23). Table 1 presents a summary of the most common risk factors.

Parental history is an underlying cause in 75%-80% of the children (24). Chorioamnionitis is a risk factor for CP, in as many as 28% of premature infants (25). Only 10%-15% of CP cases are associated with hypoxia or birth trauma.

It is reported that 60% of the children who are affected by CP are born at full term. Nevertheless, low birth weight (less than 1500g) and prematurity are well-known risk factors for CP (26-28). Perinatal risk factors include multiple pregnancies, which carries a significantly increased risk of CP in infants. Postnatal causes include head trauma, meningitis, encephalitis, and brain infarcts (20, 21). Postnatal events account for 12-21% of CP cases (23).

The range of causative factors varies at different gestational ages (i.e., pre-term, in-term and near-term infants), and in different CP subtypes (i.e., hemiparetic, diplegic and quadriparetic; *definitions are provided in section 2.1.5 Classification*). For example, the causes of hemiplegic CP, according to neuroimaging studies, are perinatal stroke and congenital malformations. In contrast, spastic diplegia, the CP subtype that is most common in premature infants, can also occur in babies born at term. Causal factors for spastic diplegia include intrauterine infection, premature rupture of membranes, and multiple gestations. Quadriplegic CP can be caused by any pathology that inflicts bilateral and widespread damage to the brain (22).

The aetiology of CP is diverse and multifactorial (23). Many of these risk factors may be part of a disease process rather than a single cause. Although prematurity and hypoxic-ischaemic injury are well-recognised contributors to the pathogenesis of CP, as many as 30% of CP cases may be genetic in nature (29, 30). Despite the small number of cases involved in current studies, there is a growing body of evidence that genetic contributions to CP are significant and important in understanding the disorder (31).

Table 1 Risk factors associated with cerebral palsy

Pre-natal	Peri-natal	Post-natal
- Iodine deficiency, iron deficiency and poor nutrition	- Birth asphyxia	- CNS infections such as viral encephalitis, tubercular meningitis and pyogenic meningitis
- Intrauterine infections, high fever, UTI	- Prematurity	- Head injuries
- Chorioamnionitis	- Intrauterine growth retardation	- Anoxia
- Hypertension	- Hyperbilirubinemia	- Suffocation
- Maternal diseases	- Intraventricular and intracerebral bleeds	- Electrocution
- Teratogens	- Hypoglycemia, dyselectrolytemias	- Postoperative cardiac arrest
- Fertility problems	- Sepsis, pneumonia and meningitis	- Post epileptic
- Poor antenatal care	- Premature separation of placenta	- Cerebrovascular accidents
- Poor socioeconomic status		- Gastroenteritis and dehydration

2.1.4 Diagnosis

Diagnosis of CP is challenging before the age of one year old, unless the condition of the child is quite severe. The diagnosis is reached through consideration of the medical history, including risk factors and genetic background, and a through physical and neurological examination (32, 33). Essential investigations include laboratory analysis (e.g., to discover any pre and postnatal infections) and neuroimaging (cranial ultrasound, computed tomography (CT), and Magnetic Resonance Imaging (MRI)) (34). Serial developmental evaluations are also necessary in the young child in order to reach the correct diagnosis and to follow-up on the child's development (32).

Early clinical signs suggestive of CP include developmental delay, toe-walking, persistent fisting, microcephaly, epilepsy, irritability, poor sucking, handedness before two years of age, and scissoring of the lower limbs. In addition, the persistence of primitive reflexes can be an early indicator. A multidisciplinary evaluation is recommended and may necessitate input from neonatology, paediatric neurology and paediatrics at the initial stage of investigation and clinical assessment (32).

2.1.5 Classification

The purpose of the classification is to describe the condition in detail since children with CP differ clinically, to predict the current and likely future prognosis of the child's condition, to compare with other children with similar type of CP and to evaluate any changes that occur after any interventions (35). The next sections offer a general overview of CP classification according to topographic, neurological and the functional classification system.

2.1.5.1 Topographic classification

CP traditionally was classified based on topography, which focuses on the distributional pattern of the affected limbs (Table 2). This classification comprises: (1) unilateral, which is used to describe hemiplegia and monoplegia, (2) bilateral, which is used to describe diplegia, quadriplegia and triplegia. *Monoplegia* and *triplegia* are relatively uncommon and are used to describe asymmetry of involvement. In most studies, diplegia is reported to be the most common form of CP (30-40% of cases), hemiplegia accounts for 20-30%, and quadriplegia accounts for 10-15% (23).

Table 2 Description of topographic classification

Classification	Description
Unilateral	
Hemiplegia	Involves one side of the body
Monoplegia	Involves one limb
Bilateral	
Diplegia	Involves both lower limbs and, to a lesser degree, the upper limbs
Quadriplegia	All four limbs are affected, the posture of the trunk, head and neck are also impaired
Triplegia	Involves three limbs, typically both legs and one arm.

2.1.5.2 Neurological classification

This allocation is based on the type/nature of motor or movement disorder (quality and changes in tone) and classifies CP into two types: *spastic,* which affects the corticospinal (pyramidal) region of the brain, and *non-spastic,* which affects the other regions of the developing brain (extrapyramidal). Extrapyramidal CP is of dyskinetic (dystonic, athetoid, choreiform), ataxic, and rigid type (19, 36). Children may develop a mix of these types. Symptoms depend on the type of CP that the child displays. The clinical features of these types are shown in Table 3. Spasticity is the most common type and accounts for 80-95% of CP cases (37).

Table 3 Description of physiological classification

Major type	Description
Spasticity	- Resistance to externally imposed movement increases with increasing speed of stretch and varies with the direction of joint movement - Resistance to externally imposed movement rising rapidly above a threshold speed or joint angle
Dyskinesia	- Abnormal movements in the arms and legs, making it difficult to control body movements and coordination
Dystonia	- A movement disorder in which involuntary sustained or intermitted muscle contraction cause twisting and repetitive movements, abnormal postures, or both
Athetoid	- Abnormal, involuntary movement - Joint contractures are uncommon - Dystonia or hypotonia can be associated
Choreiform	- Continual purposeless movements
Ataxic	- Disturbance of movement coordination, most evident in walking - Normal head/neck control - Low muscle tone and normal deep tendon reflexes
Hypotonia	- Low muscle tone and normal deep tendon reflexes
Mixed type	- Features of more than one type
Rigidity	- The resistance to externally imposed joint movement is present at very low speeds of movement, does not depend on imposed speed, and does not exhibit a speed or angle threshold - Simultaneous co-contraction of agonists and antagonists may occur, and this is reflected in an immediate resistance to a reversal of the direction of movement about a joint - The limb does not tend to return toward a particular fixed posture or extreme joint angle - Voluntary activity in distant muscle groups does not lead to involuntary movements about the rigid joints, although rigidity may worsen.

2.1.5.3 Functional classification

Currently, emphasis is placed on the functional classification due to its importance in the guidance of treatment and discussion of treatment goals (38). It also provides information that enables the evaluation of change at different points in time after any intervention (35). Thus, CP is classified according to scales that measure various functional impairments such as communication, gross or fine motor function and oromotor/oropharyngeal function. These scales are used to categorise the child's functional abilities or the severity of activity limitation and are not used as outcome measures for assessment (39). There are four common classification systems according to the type of the function that is assessed: the Gross Motor Function Classification System (GMFCS), the Manual Ability Classification System (MACS), the Communication Function Classification System (CFCS), and the Eating and Drinking Ability Classification System (EDACS) (38, 39).

Table 4 shows the levels of each classification. Each is useful in its field, but since the focus of the work described in this thesis is on ambulant children, the GMFCS is the most pertinent classification in this research and is explained in the next sections.

Table 4 Summary of the four common functional classifications

Level	GMFCS	MACS	CFCS	EDACS
I	Walks without limitation; speed, balance and coordination are limited	Handles objects easily and successfully; at most, limitations in performing manual tasks requiring speed and accuracy	Sends and receives with familiar and unfamiliar partners effectively and efficiently	Eats and drinks safely and efficiently
II	Walks with limitation, especially over long distances; on uneven terrain and inclines, and in crowded or confined spaces	Handles most objects but with somewhat reduced quality and/or speed; may avoid some tasks	Effective but slower paced sender and/or receiver with unfamiliar and/or familiar partners	Eats and drinks safely but with some limitations to efficiency
III	Walks using a handheld mobility device when indoors; uses wheeled mobility when travelling long distances	Handles objects with difficulty; needs help to prepare and/or modify activities	Effective sender and receiver with familiar partners	Eats and drinks with some limitations to safety; there maybe limitations to efficiency
IV	Self-mobility with limitations; may use powered mobility	Handles a limited selection of easily manageable objects in adapted situations; requires continuous support	Inconsistent sender and/or receiver with familiar partners	Eats and drinks with significant limitations to safety
V	Transported in manual wheelchair in all settings	Does not handle objects and has severely limited ability to perform even simple actions; requires total assistance	Seldom effective sender and receiver even with familiar partners	Unable to eat or drink safely; tube feeding may be considered to provide nutrition

Gross Motor Function Classification System (GMFCS)

This is the classification that is most widely used in the clinical and functional assessment of CP (35). The GMFCS takes into consideration a child's self-initiated movement, with emphasis on sitting, transfers and mobility. It provides a standardised system to measure the severity of movement disability. The GMFCS is a five-level, ordinal grading system that ranges from walking without restriction (level I) to the inability to maintain antigravity head and trunk posture (level V) (Table 4) (35, 38, 39). In comparison with the topographical and neurological classifications, almost all children with spastic hemiplegia can walk independently (GMFCS I); most of those with spastic diplegia can walk with or without the aid of

assistive devices (GMFCS II, III). Those with spastic quadriplegia rarely have functional walking (GMFCS IV, V) (4).

This classification was first used in 1997 by Palisano et al. (40) to describe gross motor abilities in children aged less than 12 years. The scale was revised and expanded in 2007 to increase the age range of the children to 18 years (41). It describes the skills of children in five age groups: under two, two to four, four to six, six to 12 and finally 12 to 18 years of age. These more detailed and age-appropriate descriptors enable the allocation of a more accurate classification.

This system is well regarded because it enables clinicians to compare the clinical condition of children at the same age and GMFCS level and to consider prognostic information on expected gross motor progress over time. This system is helpful to give the children and their families a prognosis for the expected gross motor progress over time; it is useful for orthopaedic surgeons to have data from wide and uniform case studies and to assist with realistic goal setting and appropriate management planning (33).

The reliability and validity of the GMFCS has been reported in several studies that show moderate to excellent inter-rater reliability and good validity (42-45). GMFCS levels remained relatively stable in children with CP (42, 43, 45, 46) and offered a valid prediction of future motor function (43, 45). Family-reported GMFCS levels have also been found to offer a reliable method for the assessment of gross motor function in children between the ages of six and 12 years (47). Therefore, not only can medical providers classify children rapidly and accurately in clinical settings through application of the GMFCS, but parents and caregivers are also effective at identifying the GMFCS level of their children.

2.1.6 Clinical manifestations

CP results from a permanent, static lesion in the developing brain that occurs before, during, or after birth (48). Even though the lesion itself does not change, its clinical manifestations alter as the child grows and develops. The clinical manifestations of CP depend on the magnitude, extent and location of the injury that caused irreversible damage to the brain, and the ability of the CNS to adapt after the insult (49). Clinical manifestations may include primary and secondary impairments.

Primary impairments are permanent, direct effects of the brain lesion, and include ‘positive’ and ‘negative’ signs (Table 5) (50, 51).

Table 5 Primary impairments

Signs	Primary impairments
Positive signs	- Spasticity - Co-contracture - Clonus - Hyper-excitability - Released primitive reflex patterns
Negative signs	- Muscles weakness - Fatiguability - Loss of selective muscle control - Poor balance and deficient equilibrium reactions - Relative imbalance between muscle agonists and antagonists across joints - Sensory problems - Cognitive impairment

Secondary impairments occur due to the consequences of primary impairments. For example, the effect of spasticity on the growth and development of skeletal muscle results in muscles that have fewer fibres, shorter fibre length, and longer tendons than muscles in children who have no spasticity. This results in a weakened muscle with a diminished excursion because it has a decreased cross-sectional area, and this, in turn, results in a decreased joint range of motion (RoM). Restriction in the RoM of affected joints leads to static muscle contractures in which the muscles are both tight and too short. This disability may lead to delayed acquisition of gross motor skills and formulation of atypical gait patterns, which lead to further deformity (4, 51).

2.1.7 Natural History

Although CP has been defined as a non-progressive brain lesion, there is deterioration in motor function and gait over time, which represents the progressive aspect of the condition (4, 49). Children who are classified as being in the lower ambulatory levels of the GMFCS are most likely to maintain their long-term function and gait. The main factor that contributes to the deterioration of function and gait is growth. Children grow rapidly and the uneven forces on their muscles, abnormal muscle tone, weakness, fatigue and contractures lead to skeletal deformity and lever arm dysfunctions (LAD). A force acting upon a lever arm attached to a fulcrum will generate torque, which is quantifiable as a moment. The amount of torque generated is proportional to the force applied and to the length of the lever arm. This is expressed by the formula M (Moment)= F (Force) X D (Distance between the force and the fulcrum) (52). Skeletal deformity affects the lever arm length and/or integrity. As a result, moment generation is affected by skeletal deformity. This contributes to the deterioration of the child's functional ability. Clinicians need

to take this into account when treatment interventions are planned. Orthopaedic surgery is one of the potential interventions and this is the focus of this project.

2.1.7.1 Gross Motor Function

Gross motor development curves have been published using the Gross Motor Function Measure (GMFM). The GMFCS was used to classify different development patterns in motor function of children with CP from birth to adulthood. Initially, the motor function curves were developed in terms of the GMFM-88, which comprises 88 measurement items. Palisano et al. (53) tested multiple mathematical models to generate best-fit curves of the data. They found that a model which assumed that children with different GMFCS levels differed in both their maximum limit of gross motor function and the rate at which that limit was approached most accurately represented the data (53). The authors noted that the predicted maximum GMFM-88 score was shown to be higher for each successively higher functional GMFCS level (53).

The results showed that, apart from level II, the curves began to plateau at around the age of seven years. This finding indicated that, by around this age, the gross motor function abilities of children with CP that were measured on the scale stopped changing. Children categorised as level II were found to approach their predicted maximum GMFM-88 score more slowly than those children categorised as level I or III. However, this finding does not indicate that CP children stop becoming more functionally advanced at this age (i.e., the children are still able to improve their ability to complete tasks that reduce the need for assistance by a caregiver, such as being able to transition from a wheelchair to a bed without assistance) (53).

After initial proof that the mathematical model was accurate enough to generate accurate gross motor function curves, GMFM-66 scores (comprised of 66 items) were used (54). Once again, the predicted limit of the GMFM-66 score increased as functional GMFCS level increased. The data indicated that children with CP who were classified as levels III, IV, and V had significantly faster rates of motor function development according to GMFM-66 score than those classified as level I. They also showed that children with CP who were classified as level II exhibited similar rates of motor function development as those who were classified as level I (54). Again, the authors emphasised that faster rates of motor development did not indicate that a child would have a higher level of gross motor function; only that the child would reach their maximum GMFM-66 score faster than children of other categories.

With the continuing development of the gross motor function curves, expected GMFM-66 maximum limits were calculated along with ranges to include 50% of the children's limits for each GMFCS level (54). For ambulant children, the GMFM-66 limit for those classified as GMFCS level I was 87.7 with a 50% range of 80.1 to 92.8%. The GMFM-66 limit for children classified as GMFCS level II was 68.4 with a 50% range of 59.6 to 76.1%. The GMFM-66 limit for children classified as GMFCS level III was 54.3 with a 50% range of 48.5 to 60.0% (Figure 1).

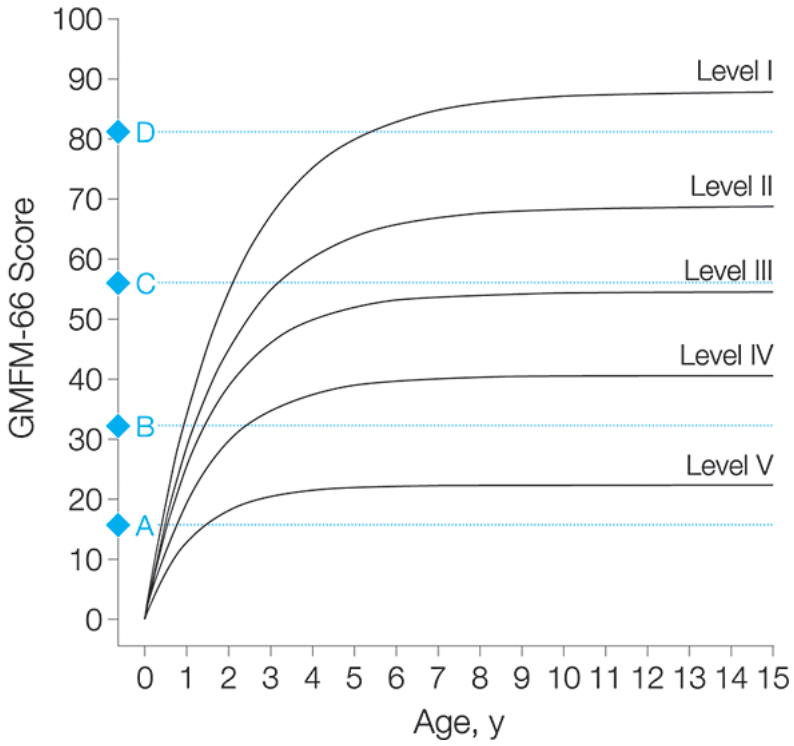


Figure 1 Predicted Gross Motor Function Measure (GMFM-66) motor scores as a function of age by Gross Motor Function Classification (GMFCS) level

In 2008, reference percentiles were created to offer approximate evaluation of a child's gross motor abilities compared with averages for the child's GMFCS level and age (55). Hanna et al. (55) used the same data set of the abovementioned gross motor function curves to create the reference percentiles. A separate percentile curve was created for each GMFCS level. The reference percentile curves for each of the GMFCS levels showed the estimated percentile curves for the 3rd, 5th, 10th, 25th, 50th, 75th, 90th, 95th and 97th percentiles with an age range from two to 12 years of age. For ambulant children, this may be seen in Figure 2, which shows GMFCS levels I, II and III in Figure 2A, 3B and 3C, respectively (55).

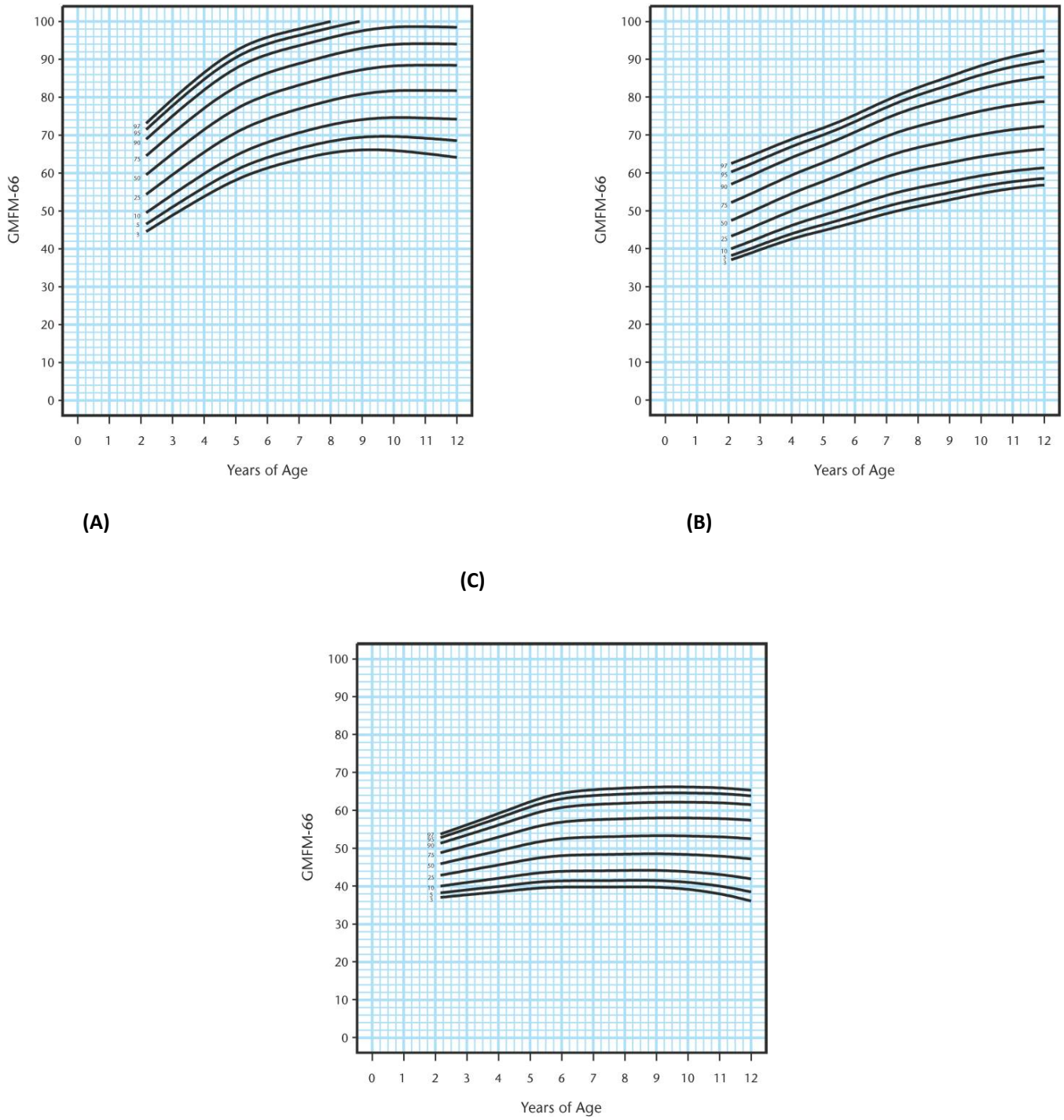


Figure 2 Gross Motor Function Classification System level (A; I, B; II, C; III) percentiles

Hanna et al. (55) also evaluated the stability of the percentiles over 12 months. For ambulant children, the mean change (and standard deviation) in percentile between multiple observations of GMFCS levels I, II, and III children were 3.0 (15.6), -0.8 (15.5), and 3.3 (12.4), respectively (55). The authors calculated the probability of a percentile change between assessments for GMFCS levels I, II and III to be 20%, 50% and 80%, and noted that the expected variability in a child's percentile over time was highest for the GMFCS levels I and II, with high variability expected for children in all GMFCS levels (55). This would

mean that the prognosis of an individual child over time does not necessarily fall within the one predicted by each GMFCS level. Authors used the same sample of the 2002 study, however, the data for each observation was treated as cross-sectional rather than longitudinal. When the authors studied the stability of the curve, they treated the data as longitudinal (54).

In 2009, Hanna et al. (56) evaluated the stability and possible loss of gross motor function during adolescence. They used the same data set from the previous study (55). The authors conducted a prospective study through calculation of the mean of five GMFM-66 motor assessments of 657 children and young adults (from 16 months to 21 years of age) with CP. The authors classified the participants according to the GMFCS and reported that ambulant children were likely to retain their mobility up to the age of 21 years as a minimum (56). The predicted average GMFM-66 trajectories from the best models in each GMFCS level are illustrated in Figure 3. However, the study findings refer to the natural history of treated children with CP with no data for untreated children. In addition, the study did not report any details of the types of treatment that the children had received and whether this could have affected the GMFCS outcome in adulthood.

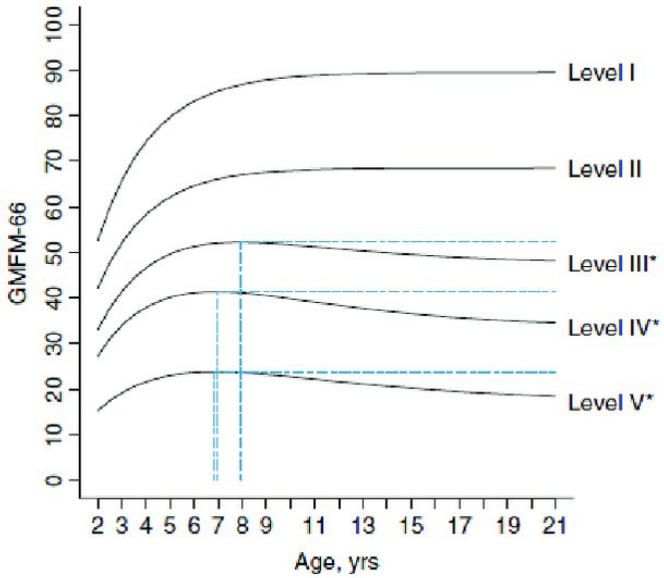


Figure 3 Predicted GMFM-66 scores as a function of age by GMFCS level. Dashed lines illustrate age and score at peak GMFM-66 . The dashed lines illustrate the timing of the peak and highlight the loss from the peak to the age of 21 years.

2.1.7.2 Gait abnormalities

Gait patterns in children with CP can be influenced by muscle weakness, spasticity, poor selective motor control, sensory abnormalities, and secondary musculoskeletal changes. Gait is a complex motor task

that requires coordination of many degrees of freedom. Gait analysis is the systematic study of human locomotion. It may include simple observation using scoring systems; video recording and two-dimensional analysis; and instrumented three-dimensional analysis using advanced opto-electronic technology, as described in a later section: 3.1.2 Assessment of lower limb orthopaedic surgery. Instrumented clinical gait analysis usually involves a comparison of impaired or pathological gait with normative gait curves from peers who develop in a typically manner. It is an important element of assessment in ambulant children with CP, since gait reveals valuable information that is relevant to the diagnosis and treatment of musculoskeletal disorders. Therefore, it is necessary to firstly understand the characteristics of typical function and walking patterns in individuals without gait impairments.

Normal gait

Human gait is dependent on several factors that include adequate function and motion of the skeleton, enabled by balanced muscular activity, which is initiated and modified by the CNS. These factors evolve through childhood. During normal development, a mature gait pattern is established by the age of four to five years, which means that *kinematic* movements (between body segments) and *kinetic* forces and moments (which produce the movements) produce gait variables that are within adult ranges (57). *Temporal- spatial parameters*, which include step and stride length cadence and gait speed, are influenced by other factors, e.g., body height and leg length. Therefore, these measures do not reach fully mature values until growth is complete (57, 58). Kinetics comprise *internal forces* in muscles, joints and ligaments, and *external forces* such as the Ground Reaction Force (GRF). The GRF comprises vertical components due to gravity, and horizontal shear forces that are caused by friction with the ground. When a force acts at a distance to the joint axis of rotation (fulcrum), the perpendicular distance of the force vector from the fulcrum is called the *lever arm*, and the product is the *joint moment* (which is usually measured in Newton metres, Nm) (59). For a joint to flex or extend, an appropriate moment should be generated.

Gait variables are typically analysed using the gait cycle as the measurement unit. A gait cycle starts with initial contact of one limb with the ground, includes the stance (60%) and swing periods (40%) and ends with the subsequent initial contact with the same limb (100%). One *stride* comprises two successive steps with the right and left limbs. The *step length* is the distance from the toe of the trailing foot to the point of contact of the opposite leading foot. The number of steps or gait cycles made per time unit is referred to as the *cadence* or *frequency*. During stance, about 20% of the cycle involves double support in which both feet are on the ground simultaneously.

Researchers have defined the 'five priorities of normal gait' which summarise the main tasks of functional human walking, including (1) stability in stance, (2) foot clearance in swing, (3) pre-positioning of the foot, (4) adequate step length, and (5) energy conservation (60). These priorities may be helpful in the identification of deviations and in understanding the main challenges in impaired gait.

Gait patterns in CP

Gait in CP can be classified into distinct patterns through the use of classifications that combine visual observations and quantitative, kinematic data from three-Dimensional Gait Analysis (3DGA) (61, 62).

Rethlefsen et al. (63) evaluated the prevalence of gait deviations in more than 1000 patients with bilateral and unilateral spastic CP who were classified as GMFCS levels I to IV. The study confirmed that, primarily in children who could walk independently or with a minimum support (levels I-III) and only in children with bilateral CP, increased age brought an increased risk of crouch, which is characterised by excessive dorsiflexion at the ankle in combination with excessive flexion at the knee and hip joints (61). The prevalence of equinus gait; which is defined by the ankle in plantarflexion throughout stance phase and the hips and knees extended (61); and in-toeing was found to decrease with age and higher GMFCS levels (63). The authors recommended caution among physicians who made treatment decisions for younger children, due to the chance that equinus and in-toeing would decrease naturally with age.

Bell et al. (64) investigated changes in gait in children with diplegia who had not had surgery. They found that children with a mean age of 7.8 years at the start of the study experienced deterioration in gait over a 4.4-year period (mean value). More specifically, elements of the gait cycle such as stride measures, passive range of motion and kinematic parameters deteriorated. However, the researchers did not compare results for the study participants with the results for those who had surgery (64).

2.1.8 Clinical management

To date, there is no treatment to reverse the pathology that underlies CP, i.e. the brain injury that causes the disorder. Therefore, clinical management focuses on the management of the clinical manifestations, such as musculoskeletal abnormalities (e.g., muscle stiffness, muscle contracture, joint deformity and torsional deformities) (65). Individuals with ambulatory CP typically experience deterioration of function and gait as they grow into adolescence, therefore management is primarily designed to prevent functional loss and to improve or optimise gait (66).

A number of intervention strategies are used successively or in combination. These include physiotherapy; orthotics (braces) or serial casting; pharmacological or neurosurgical methods; and orthopaedic surgery.

Physiotherapy, which comprises exercises that are used to: (1) prevent or reduce muscle and joint contractures; this is achieved by passive or dynamic range of motion exercises and stretches across major joints (e.g., ankle, knee, hip); (2) increased muscle strength is achieved by performing progressively challenging resistance exercises involving all major muscle groups (e.g., gastrocnemius, rectus femoris, hamstring muscle, and hip flexor); and (3) low resistance, high repetition exercises of major muscle groups to improve local muscular endurance. In addition, there are other specific physiotherapy interventions that are designed to improve balance, postural control, gait, and assist with mobility and transfers (67, 68).

Application of orthotics (braces) or serial casting, which are "externally applied device(s) used to modify the structural and functional characteristics of the neuro-muscular and skeletal systems" (69). The Ankle-Foot Orthosis (AFO) is the most common and typically prescribed orthosis for ambulant children with CP, with the purpose of maintaining range of motion, preventing deformity, improving the base of support in both standing and functional walking through enhanced stability in stance and assisting foot clearance and prepositioning for initial contact (70).

These methods are often accompanied by measures to reduce muscle tone by *pharmacological* (e.g., oral medications or through intra-muscular injection of botulinum toxin-A (BTX-A)) or *neurosurgical methods* (e.g., selective dorsal rhizotomy (SDR)). Clinicians believe that these methods facilitate the muscle stretching that is undertaken through therapy and serial casting and improve tolerance of brace wearing, which in turn may prevent or delay the onset of contractures and bone deformities. However, musculoskeletal deformities can occur despite those preventative interventions and are commonly addressed through *orthopaedic surgery* (48, 51, 66). Orthopaedic surgery for CP is complex and resource-intensive, it requires significant time and effort from children and their families and involves extensive rehabilitation.

2.1.9 Orthopaedic surgery in cerebral palsy

2.1.9.1 Indications and goals of surgical interventions

The goals of orthopaedic surgery depend on the severity of the condition, functional impairment, and level of ambulation according to the GMFCS level. These goals are defined by the child with CP, their family, and the multidisciplinary team. The goals that are usually set when undertaking orthopaedic surgery on ambulatory children with CP are to treat or prevent pain, preserve or improve physical function, and optimise gait efficiency (71).

The objectives of surgery facilitate planning for lower limb orthopaedic surgery. For instance, correction of Lever Arm Dysfunction (LAD) is a common objective of interventions that are used to improve gait for children with CP, especially in orthopaedic surgery and orthotics design. Unstable LAD can be corrected by orthopaedic surgery irrespective of age (72, 73). Sharan (73) argued that the best functional outcomes are achieved before severe LAD and joint decompensation sets in. The risk of recurrence of contractures is minimised by correction of the LAD. Multiple clinical studies have shown that surgical interventions to correct femoral anteversion, which is the inward rotation of the femur bone in the upper leg, considered as one of the most common sources of LAD among children with CP. Derotation osteotomy can correct internal hip rotation during gait in the short- to mid-term (52, 74). However, Steele et al. (74) reported that there was no evidence on the consequences of treated and untreated LAD on long-term function and Quality of Life (QoL). They noted the need for additional evidence to understand these issues and provide options and guidance for adults with CP.

The presence of significant hip displacement is an indication for orthopaedic surgery, irrespective of age. Robb et al. (75) stated that operative prevention should be considered in children with a Reimers' migration percentage (MP) that exceeded 33%. Reimers' MP is the most commonly used, validated and reliable measure for hip displacement (76). A surveillance programme that is based on the child's age, GMFCS level and MP is important to identify children who are deemed at risk of hip displacement, to monitor their hip development over time, and to offer early appropriate intervention (75, 77). Studies have found that most hip dislocations are observed in GMFCS levels III, IV, or V (78-81).

The role of Instrumented Gait Analysis (IGA) has been noted as a preoperative evaluation, as it significantly affects decision making and surgical indications for orthopaedic surgery. For example, the surgical team may decide to include surgery in the context of femoral derotation osteotomy, if gait

analysis data highlight the presence of femoral anteversion (82). Several studies show that surgeons follow recommendations that are made after performance of IGA 42-97% of the time (83, 84). In cases where surgery has already been decided, the addition of IGA has been shown to lead surgeons to change 40-89% of their surgical treatment plans (83, 85-87). The investigators of these studies were not blinded, their decisions were based on observation alone and there was an absence of a control group. Nevertheless, the IGA is considered a valuable tool in clinical decision-making, and an optimal preoperative evaluation through IGA is a prerequisite for multi-level surgery. IGA provides valuable information after surgery to assess the operation's outcome (82, 83).

Recently, international consensus-based studies have been undertaken to determine the indications and parameters that are taken into account during surgical decision making for medial hamstring lengthening (MHL), femoral derotational osteotomy (FDO) (88), gastrocnemius/soleus lengthening (GSL) (89) and distal rectus femoris surgery (90) in ambulatory children with CP. The studies involved the participation of expert orthopaedic surgeons. In McCarthy et al.'s 2020 study (88), experts agreed on ten indications for MHL and 29 indications for FDO including clinical examination, imaging and gait analysis parameters. Rutz et al. (89) conducted three rounds of a Delphi study with 17 experienced orthopaedic surgeons to identify indications for GSL surgery. They took into account the child's age, CP type, clinical and laboratory examinations, and goals. They reached consensus thresholds (80% agreement) on eleven indications and goals. In Kay et al.'s 2021 study (90), experts agreed on 17 of 33 indications regarding distal rectus femoris surgery through a Delphi consensus process. From the 17 statements, 12 indications reached consensus in accordance with the study's agreed level (at least 80%), one against the statements and four reached general agreement level (60%-79%). Similar to McCarthy's study, indications included parameters related to general characteristics, clinical indications, gait analysis data, intraoperative techniques and outcome measures.

2.1.9.2 Nature of the procedure

In ambulant CP, orthopaedic procedures to address the musculoskeletal deformities of the lower limb include the lengthening of musculotendinous units, tendon transfers, osteotomies and arthrodesis. Orthopaedic surgery should be considered to correct deformities in all three planes (sagittal, axial and coronal) at the levels of the hip, knee, ankle and foot.

In the *sagittal plane*, spasticity and muscle imbalance lead to contracture of the muscle-tendon units (MTUs) on one side of the joint. *Axial plane* problems are torsional abnormalities, with femoral anteversion and external tibial torsion being the most common. *Coronal plane* abnormalities predominantly affect the foot and ankle; with *equinoplanovalgus*, which is characterised by hindfoot and/or forefoot valgus/pronation or midfoot break in stance or swing, with or without equinus, or *equinocavovarus,* defined by the static and dynamic imbalance of the agonist and antagonist muscles that control the foot and ankle joints causing hindfoot varus, midfoot cavus and forefoot equinus. These are the most common deformities in bilateral and unilateral disease respectively.

Hip

Fixed Flexion Deformity (FFD) of the hip is a sagittal plane problem that prevents full hip extension. The GRF is moved posterior to the knee due to the flexion of the hip and the position of the trunk. In the axial plane, the femur commonly exhibits abnormal torsion, with excessive femoral neck anteversion, which contributes to internal hip rotation and can lead to a rotational LAD. In the coronal plane, the femoral neck is sometimes in valgus; an increase in the angle between the femoral neck and the femoral diaphysis; and, in some cases (depending on GMFCS level), the hip is displaced.

Surgical management of the torsional femoral deformity involves a femoral derotation osteotomy (FDRO). FDRO can be performed proximally (inter-trochanteric) or distally (supracondylar). The most commonly used method involves a proximal metaphyseal osteotomy and fixation with a fixed-angle plate. A coexisting FFD at the hip or knee can be corrected by fixing the osteotomy in compensatory extension.

Knee

Orthopaedic procedures for this joint comprise various combinations of musculotendinous lengthening of the distal hamstrings, transfer of the distal hamstrings, lengthening or transfer of the rectus femoris, external rotation and/or extension osteotomy of the distal femur, distalisation of the patella and surgery to the distal femoral growth plate (known as ‘guided growth’ surgery).

FFD of the knee is a sagittal plane deformation that can lead to a positional LAD and displacement of GRF posterior to the knee. Short hamstrings may prevent full extension at initial contact. Over-lengthening or weakness of the quadriceps may prevent full knee extension in stance. Medial Hamstrings Lengthening has been widely employed and can improve knee extension; however hamstrings weakness

can result from this and lead to anterior pelvic tilt. Distal femoral extension osteotomy has become more popular in recent years for this reason. Patellar tendon shortening or distal transfer of its tibial attachment are usually combined with this procedure to restore adequate tension of the extensor mechanism. Distal rectus femoris lengthening or transfer to the medial hamstrings is used to compensate for stiff-knee gait in the swing phase of gait.

Foot and ankle

The common foot deformities are equinus, equinoplanovalgus and equinocavovarus. Deformities of the foot often affect all three planes. These can lead to the development of a short, flexible lever arm that is a major contributor to abnormal gait. Foot deformity causes instability in stance and clearance problems during swing.

Gastrocnemius contracture leads to equinus positioning of the ankle, which is a very common deformity in the sagittal plane. Children with spastic CP and equinus gait have longer Tendo-achilles and shorter muscle bellies than normal. Gastrocnemius contracture, which is commonly seen in diplegia, is generally treated through a soft-tissue lengthening of the gastrocnemius aponeurosis in the gastroc-soleus musculo-tendinous junction. In hemiplegia, the soleus also develops contractures and is treated by combined distal lengthening of the conjoined Achilles tendon.

The equinoplanovalgus deformity begins with the lateral displacement of the navicular, which causes the talar head to become uncovered and prominent in the medial mid-foot. Eversion/valgus deformity of the hindfoot and abduction of the forefoot contribute to the deformity. Lateral calcaneal lengthening with concomitant peroneus brevis lengthening and shortening or reefing of the medial foot structures is the recommended procedure to correct an equinoplanovalgus foot deformity. Medial displacement calcaneal sliding osteotomy is the preferred surgery for hind-foot valgus when the subtalar joint is no longer flexible. Subtalar or triple arthrodesis may be necessary in more advanced or neglected deformities.

Flexible equinovarus deformity can be corrected through soft tissue lengthening and tendon re-balancing surgery (e.g., tibialis anterior tendon transfer). Severe rigid equinocavovarus deformity can be treated through a one-stage correction that comprises soft-tissue surgery, combined with joint preserving osteotomies or joint fusions, depending on the severity and rigidity of the deformity.

Single-Event Multi-Level Surgery (SEMLS)

Historically, orthopaedic surgery to correct secondary musculoskeletal deformities was performed in a staged manner to address one deformity at a time, a practice referred to as 'birthday syndrome'. This meant that children often had to undergo surgery every one or two years. This resulted in unpredictable outcomes and recurrence of the deformity, as well as a need for annual surgeries, casting and physiotherapy. It was eventually found that it was necessary to correct associated deformities during correction of the deformity of initial concern; otherwise, the corrected deformity would recur (91). As a result, during the 1980s, evolving evidence indicated a shift in practice to the performance of multiple surgeries in one sitting. This is now commonly referred to as Single-Event Multilevel Surgery (SEMLS). Multi-level surgery is defined as the combination of a minimum of two operations undertaken bilaterally at a minimum of two levels (e.g. hip and knee) (92).

Multi-level surgery is most commonly guided by the findings of 3DGA (93, 94). It enables surgeons to correct both soft-tissue and bony deformities in the lower limb in a single operation (95). According to Thomason et al. (92), the principle behind the performance of an SEMLS is to achieve sagittal and axial plane balance, which should result in improved gait pattern and functioning. The advantages are that there is the need for only one hospital admission and one rehabilitation period, and secondary deformities which otherwise can occur if only one abnormality is addressed at a time are prevented (66, 96).

SEMLS requires careful planning and execution, but the rehabilitation stage is equally important. The patient loses up to 50% of their preoperative strength post-operatively (51), and if the child and family do not cooperate in the performance of the required intensive physiotherapy, the result of the operation may be poor.

Adverse Events (AEs)

Lee et al. (97) in 2015 reported the complications potentially arising from lower limb orthopaedic surgery in children with CP. They found that 2.3% of children had major and 4% had minor complications. It was reported that complications of orthopaedic surgery could be classified as under-correction, over-correction or recurrence following successful operation (4). Other complications that were reported included pain, prolonged joint stiffness beyond four weeks, pressure ulcers, fracture, wound dehiscence and infection.

2.1.9.3 Timing of orthopaedic surgery

The timing is determined by several factors such as CNS maturation, ambulation potential, and the rate at which the contractures and LAD are developing. As mentioned in sections 2.1.9, the presence of significant hip displacement is an indication for orthopaedic surgery. In the absence of hip displacement, it is recommended that any surgical intervention should be delayed until the child is at least seven to nine years old because at younger ages there is a high risk of recurrence (98, 99). Unstable LAD requires orthopaedic surgery irrespective of age in order to preserve ambulation (72, 100). Rang (101) in 2002 recommended that orthopaedic surgery should be completed between the ages of four to eight years in children with spastic diplegia. Orthopaedic surgery in adolescence is associated with an increased incidence of postoperative psychological problems and sudden disruption of the young person's body image (102).

Bache et al. (103) described a logical algorithm for the management of musculoskeletal pathology for children with spastic diplegia. The authors defined two periods of interventions that were required. The first was a period of spasticity management, from two to six years old. The second period was surgery for contracture and bony torsional problems, to be carried out between the ages of 7 and 11 years. The majority of children would undergo SEMLS to correct deformities and improve gait (103).

Quinby et al. (104) supported use of this algorithm and emphasised that orthopaedic surgery for ambulant children should be avoided before the age of seven years, since the mature gait pattern develops at that age. They stated that bony surgery for torsional correction should be undertaken after the age of eight years, as prior to this, there was a high risk of recurrence of the deformity.

A recent study conducted by Rutz et al. (89) included expert orthopaedic opinion through the use of a Delphi technique to establish consensus for surgical indications for GSL in ambulatory children with CP. Agreement was reached that the optimum age was six to ten years and that it should be avoided in children under four years of age.

2.1.9.4 Rehabilitation and recovery

It is known that a lengthy period of rehabilitation is required to reach a new functional plateau. Peterson et al. (51) noted that rehabilitation was vital; intensive rehabilitation could be required for up to two years after major surgery such as SEMLS. The researchers emphasised that without good compliance in

rehabilitation, surgery could make children with CP weaker and stiffer than they were before surgery. In adolescence, longer post-surgery recovery times are expected because teenage bones do not heal as fast as young children's following osteotomies (102). One of the major goals of post-surgical rehabilitation for ambulant children is to promote independent mobility, which includes walking. The rehabilitation process includes appropriate bracing, strengthening and gait training.

There is evidence that SEMLS followed by intensive rehabilitation is beneficial (105). Akersted et al. (106) in 2010 found that intensive training post-surgery improved children's walking ability (106). Patikas et al. (107) also found a resistive exercise programme helped to prevent muscle weakness during the postoperative rehabilitation period. However, regarding the increased weakness and stiffness that can result when there is no intensive rehabilitation or compliance with the exercise, (51, 108) Seniorou et al. (109) compared the efficacy of progressive resistance strengthening versus active exercise in children with spastic diplegia. The authors found that, 6 to 12 months after the operation, the strength of all muscle groups was significantly reduced compared with before the operation, despite improvement in gait kinematic measures. After six weeks of intensive physiotherapy, both groups showed significant improvement in muscle strength, GMFM scores and gait parameters.

Several different physiotherapy interventions are used after orthopaedic surgery for children with CP, so it is difficult to determine which techniques are optimal. A review by McGinley et al. (110) in 2012 concluded that "there is little information in the available literature to guide clinicians and scientists as to the optimum post-SEMLS rehabilitation". They reported that a protocol for post-SEMLS rehabilitation was found to be lacking in 50% of studies, despite widespread acknowledgement of its importance. A more recent review conducted in 2019 by van Bommel et al. (108), which aimed to determine the most common practice with regards to rehabilitation post-SEMLS, concluded that there was an incomplete description of the physiotherapy and variety of treatments in all studies, with a small level of agreement regarding the goals of the physiotherapy treatment and the frequency, content and intensity of training sessions.

2.1.9.5 Evidence for orthopaedic surgery

The majority of published studies on lower limb orthopaedic surgery involved small sample sizes and were of retrospective design. There were eight randomised trials (84, 111-117) in this field. Two of those

included randomisation of surgeons who received gait analysis data prior to surgery rather than randomising children to different surgical interventions (84, 117).

Dreher et al. (118) noted that surgery could have poor long-term outcomes as measured by various gait parameters. A ten-year retrospective study of ambulant children with CP who had undergone SEMLS including hamstring lengthening found that there was a significant deterioration in several gait parameters. Short-term findings had shown significant improvements in gait parameters, which was consistent with other research that had investigated short-term outcomes (119).

Nicholson et al. (120) in 2018 evaluated the relationship between the Gait Deviation Index (GDI) and walking activity pre- and post-operation in a sample of 72 ambulant people with CP. The GDI was calculated through the use of 3DGA and walking activity was monitored using the StepWatch (SW). The SW is a wearable device that monitor the steps as form of stride counts of one leg (121). Participants undertook pre- and postoperative gait analysis. The participants were monitored for 26 months after surgery. The results showed that SEMLS improved gait deviations in children with CP. However, the improvement in gait pattern showed a limited correlation with postoperative changes in walking activity (120).

Salami et al. (122) in 2019 reported on the outcomes of a sample of 19 ambulant children with CP (mean age nine years). All participants underwent hamstring lengthening as part of SEMLS; however, the report contains no clear information about the associated surgery. Follow-up assessments that investigated changes in hamstring muscle-tendon length and lengthening velocity were undertaken after one and at least four years. The results showed that the surgery was efficient only in the short term with regards to changes in muscle-tendon length. However, it did not change hamstring lengthening velocity. The reported outcome of the study may have been influenced by other surgery that was performed during the SEMLS, even though the hamstring lengthening was considered to be a central part of the operation for all the children (122).

Gorton et al. (123) conducted a well-designed prospective cohort study to compare the outcomes of children with CP who underwent SEMLS with those of children who continued with regular physiotherapy. A total of 75 patients underwent surgery and were matched by age, gender and GMFCS level to participants who did not undergo surgery. Significant improvement was reported in the children who had SEMLS at the one-year follow-up with regards to gait, as measured by the Gillette Gait Index

(GGI), and QoL, as measured by the Paediatric QoL Inventory (PedsQL) in the gait domain. No significant change was noted in the group of children who received only standard physiotherapy. The authors noted that although improvements were noted in body structure and function, there was no evidence that these would result in improvement at the activity and participation level of the ICF-CY framework (123). The conclusions that were reached corroborate other studies, which show that there is improvement in gait and QoL one year after SEMLS in comparison with children with CP who receive only physiotherapy (92, 124). However, a one-year follow-up period is insufficient to determine whether SEMLS results in a significant change at the activity and participation level of the ICF-CY framework.

Thompson et al. (92) in 2013 conducted a five-year, prospective follow-up study on 18 young people with CP. The participants were reassessed at one, two and five years after surgery. Improvements were noted at the one-year follow-up period with regards to gait parameters, and at the two-year follow-up concerning motor function and participation in motor activities. They recorded an improvement of 3.3% on the participants' GMFM-66 scores at the five-year follow-up, in comparison with the preoperative scores, as well as an improvements in the Functional Mobility Scale (FMS) over all three distances that are measured in the scale (92). When looking at a follow-up period of this length, it is essential to acknowledge the natural development of gait and function in children with CP, as described in section 2.1.7, which follows the pattern of a plateau and then a decline (64). It can therefore be concluded from this study that, in this cohort, there was improvement in gait, motor function and participation post-SEMLS two years after surgery, and that this improvement was maintained after five years.

Edwards et al. (125) in 2020 reported a long-term prospective study of 61 participants (mean age 11.8 years). The researchers aimed to study functional mobility in participants with diplegic CP at a mean of eight years follow-up after SEMLS. The participants' postoperative functional mobility was assessed using the FMS; however, this was not observed by the researchers, who completed a questionnaire by telephone. The study noted improvement in the postoperative FMS data. However, the report did not offer a comparison with preoperative data, so the study was inconclusive (125).

Well-designed RCTs that follow-up participants over the short term supported good outcomes after SEMLS in children with ambulant CP. Thomason et al. (116) carried out a pilot RCT in 2011. The trial was a single-centre pilot project to determine the sample size needed for a multi-centre study. Nineteen children were enrolled into the pilot study: 11 children were placed in the intervention group (SEMLS) and eight in the control group. The follow-up period was only 12 months, after which the control group

also received surgery. The Gait Profile Score (GPS) and the GGI, as described in section 3.1.2, were used to analyse the effects of SEMLS on gait. The FMS, GMFM-66 and time that the children spent in upright position were used to assess the functional outcome. The Child Health Questionnaire (CHQ) was used to assess each child's QoL (116).

The researchers found that the surgery group showed significant improvement in gait after the 12-month follow-up period as compared with the control group. There were no significant changes in the control group after the 12 months, despite physiotherapy that included a 12-week resistance strength programme. However, there was no significant improvement in functional ability or QoL in either group at the 12-month follow- up (116).

Klotz et al. (114) in 2018 followed up for one year 20 children who had participated in the researchers' RCT study. The study was intended to investigate: (1) whether patellar tendon shortening (PTS) improved maximum knee extension and reduced knee flexion contracture in children with flexed knee gait; and (2) whether PTS influenced the range of motion of the knee during the swing phase. Improvements were noted at the one-year follow-up with regards to maximum knee extension, reduced knee flexion, knee flexion contracture and the popliteal angle in the intervention group (SEMLS +PTS). The study noted that the findings were limited because of the small sample size and short follow-up period.

Despite the reports on beneficial objective outcomes of lower limb surgery, the improvements may not be advantageous for the children and their families. The majority of studies did not report the views of the children or their families on the benefits they expected or experienced from the surgical interventions. This is further discussed in sections 3.2 Children's expectations, preferences and priorities and 3.3.3 Unmet expectations, irrelevant outcomes.

Chapter 3 Evaluation of Surgical Outcomes

This chapter provides an overview of the literature relevant to the evaluation of lower limb orthopaedic surgical interventions on children with CP and the relevant outcome domains and outcome measures. It includes a description of the World Health Organization (WHO) International Classification of Functioning, Disability and Health- Children and Youth (ICF- CY) Framework.

3.1.1 ICF-CY Framework

3.1.1.1 Origins of the ICF-CY framework

In 1980, the WHO issued the International Classification of Impairments, Disabilities and Handicaps (ICIDH), which is a model for the classification of the consequences of disease or injury and their implications to the lives of individuals (126). The link between impairment, disability and handicap is shown in Figure 4. The ICIDH concepts provide the key to rational management of chronic diseases; however, it has been criticised as a disease-based framework. This implies a linear progression through a fixed sequence, which considers medical factors to determine outcomes. Additionally, it understates the role of the social and physical environment in the process of handicap assessment (127).

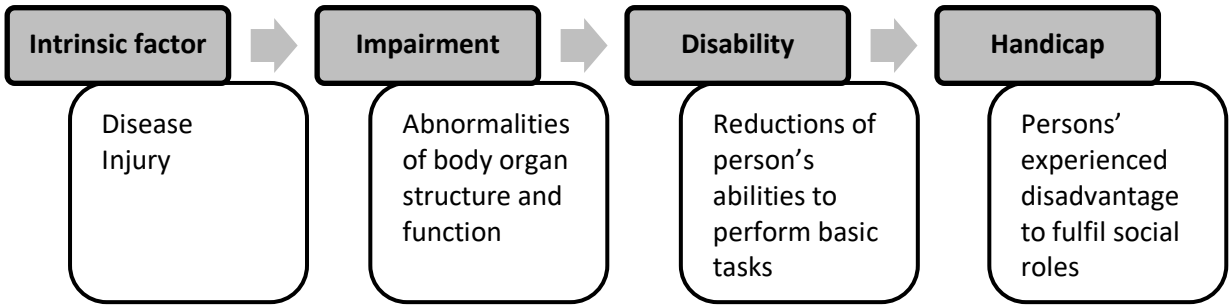


Figure 4 The International Classification of Impairments, Disabilities and Handicaps

As a consequence, an international collaboration was formed in early 1990 to address these issues and to revise the model. The newly introduced model was published in 2001 (128). The 2001 ICF framework is based on a bio-psycho-social model, which was first proposed by Engel in 1977 (129). This model provided a framework to integrate the biological, psychological and social domains in order to define health and disability. In the biopsychosocial model, *disability* is considered to involve an interaction of the individual with a particular health condition and their community or society (129).

The WHO ICF framework provides a standard language to describe health and health-related states. It helps to describe changes in a person's body functions and structure, activity and participation levels, what a person with a health condition can do in a standard environment (capacity), and what they actually do in their usual environment (performance). These components are classified as body functions and structure, and activity and participation. The ICF framework proposes that contextual factors influence and modify other components such as level of activity and participation, and must be considered during the assessment of the functional performance of individuals with a health condition (128). In ICF, the term *functioning* refers to all body functions, activities and participation, while *disability* is similarly an umbrella term for impairments, activity limitations and participation restrictions (128). Figure 5 highlights the dynamic aspect of the framework and the interaction between its components (128).

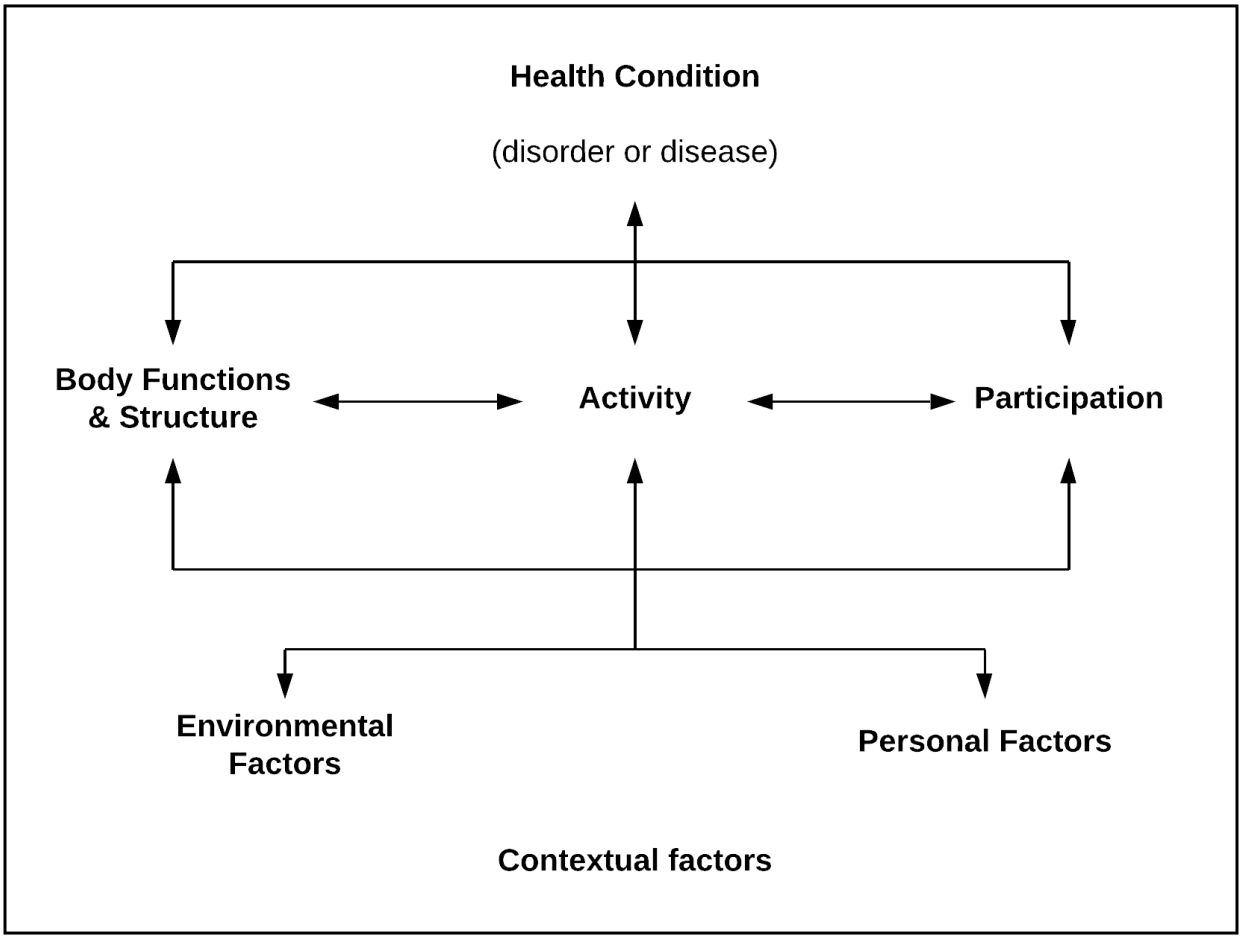


Figure 5 International Classification of functioning, disability and health

Despite the usefulness of the ICF framework, its sensitivity to the characteristics of functioning in children was limited because the manifestations of functioning, disability and health conditions in

childhood and adolescence are different in nature, intensity and impact from those in adults (130, 131). As a consequence, in 2007, the WHO issued the ICF, Disability and Health for Children and Youth (ICF-CY) (12).

The ICF-CY builds on the ICF; it adds domain descriptions to encompass developmental aspects relevant to children and young people (from birth to 18 years). The most significant changes are in the activity/participation component (132). For instance, the ICF-CY mobility category includes "scooting and rolling" and "shuffling" items, which were not part of the original ICF framework.

Furthermore, in the ICF-CY framework, the term 'developmental delay' and its concept were added to define the universal qualifier for body functions and structure and activities and participation levels due to the differences in growth, development and the acquisition of skills in children. Lag in the emergence of functions, structures or capacities may not be permanent but reflect delayed development (12).

3.1.1.2 Overview of the ICF-CY framework

As in the ICF framework, the classification of the ICF-CY is organised into two parts and three main components: the body (function and structure); activities and participation; and contextual factors (environmental and personal) (12, 128). A description of each component is shown in Table 6.

Table 6 ICF-CY Framework

ICF-CY Part	ICF-CY Components	Description
Part 1: Health status		
	Body function	The physiological and psychological working of body systems
	Body structure	Anatomical parts of the body, such as limbs and their components
	Activity	The execution of a task by an individual
	Participation	The extent of a person's involvement in different areas of life and range from basic skills
Part 2: Contextual factors		
	Environmental factors	Physical, social, cultural or institutional in nature
	Personal factors	Gender, age, level of education and lifestyle

The activity and participation component is made up of a single list that covers the full range of daily life areas. The component can indicate activities (a) or participation (p) or both. The domains of this component are bounded by the two qualifiers of capacity and performance. *Capacity* describes an

individual's ability to execute a task or an action (activity), while *Performance* describes what an individual does in his or her current environment (participation). However, it is difficult to distinguish between activities and participation on the basis of the categories in this component. The WHO has suggested that some domains should be designated as activities and others as participation, with overlap prohibited (12).

The difficulty in differentiating between activity and participation has been noted in the literature. Adolfsson (133) stated that additional work was required to create reliable and valid instruments that made it possible to discriminate between the two.

3.1.1.3 Alphanumeric codes of the ICF-CY framework

In the classification, letters and numbers are used to indicate different ICF-CY levels. The components of body functions, body structures, activities and participation and environmental factors are represented as letters b, s, d, and e, respectively. The letters are followed by a code number, which indicates a chapter number (first level) to represent a broad area of functioning, followed by codes for the second, third and fourth levels which provide greater specificity regarding the area of functioning (12, 132). For example, a child who requires assistance for mobility would be assigned the code d4501: d4 for mobility (chapter, 1st level), d450 for walking (2nd level), and d4501 for walking long distances (3rd level) (Figure 6). Figure 7 shows the distribution of the ICF-CY categories by components.

Components of the ICF-CY are quantified using a generic scale that ranges from 0 (no problem) to 4 (complete problem). These scale components are termed 'qualifiers'. Body functions are coded by one qualifier that indicates the extent or magnitude of the impairment (i.e., deviation, loss or delay). Body structures are coded by three qualifiers: extent of impairment, nature of impairment and location of impairment (Table 7). Activity and participation are coded by two qualifiers (i.e., performance and capacity), while environmental factors also have two qualifiers: the negative aspects of the environment are qualified in terms of barriers, whereas positive aspects are denoted as facilitators (Table 8) (12, 132).

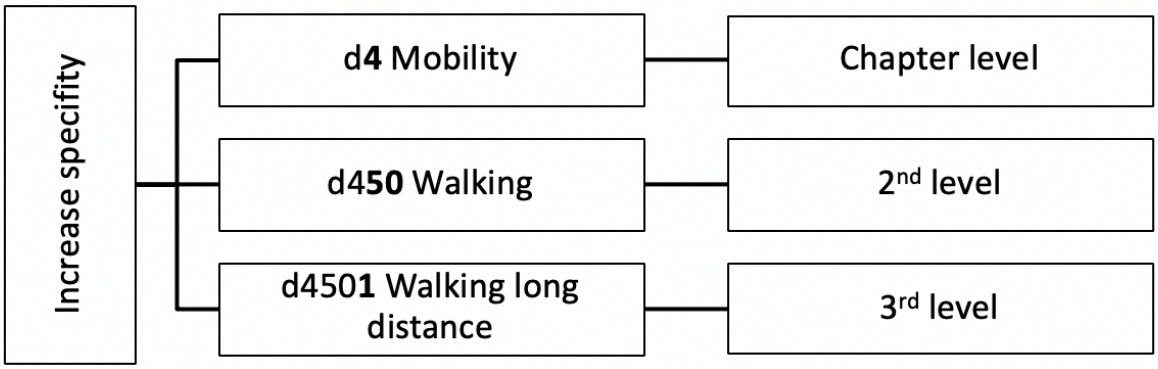


Figure 6 Example of Alphanumeric Codes of ICF-CY Framework

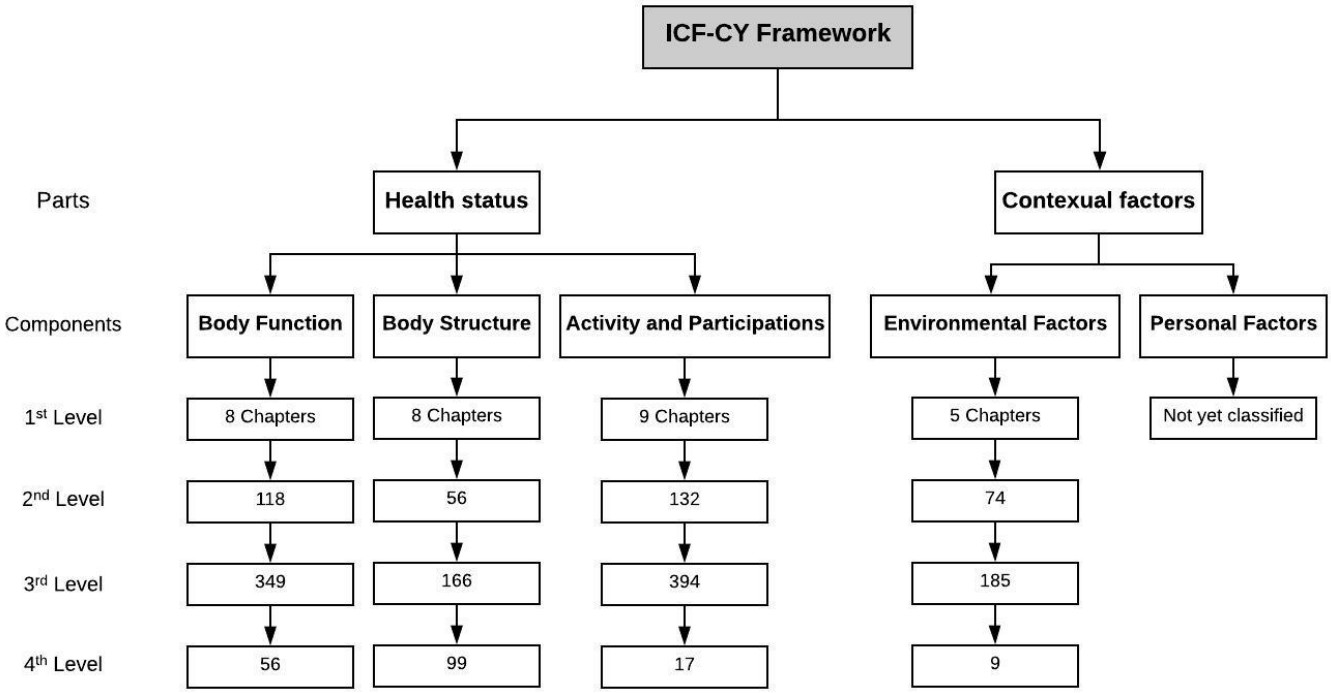


Figure 7 Structure of the ICF-CY Framework

Table 7 ICF-CY qualifiers for the body functions and body structures

Body Function	**Body Structure**		
First Qualifier 'Extend or magnitude'	**First Qualifier** 'Extent or magnitude'	**Second Qualifier** 'Nature'	**Third Qualifier** 'Location'
0 No problem	0 No problem	0 No change in structure	0 More than one region
1 Mild problem	1 Mild problem	1 Total absence	1 Right
2 Moderate problem	2 Moderate problem	2 Partial absence	2 Left
3 Severe difficulty	3 Severe difficulty	3 Additional part	3 Both side/ median
4 Complete difficulty	4 Complete difficulty	4 Aberrant dimensions	4 Front
8 Not specified	8 Not specified	5 Discontinuity	5 Back
9 Not applicable	9 Not applicable	6 Deviating position	6 Proximal
		7 Qualitative changes in structure, including accumulation of fluid	7 Distal
		8 Not specified	8 Not specified
		9 Not applicable	9 Not applicable

Table 8 ICF-CY qualifiers for activity and participation and environmental factors

Activity and Participation		**Environmental Factors**	
First Qualifier 'Performance'	**Second Qualifier** 'Capacity'	**First Qualifier** 'Barrier'	**Second Qualifier** 'Facilitator'
0 No problem	0 No problem	-0 No barrier	+0 No facilitator
1 Mild problem	1 Mild problem	-1 Mild barrier	+1 Mild facilitator
2 Moderate problem	2 Moderate problem	-2 Moderate barrier	+2 Moderate facilitator
3 Severe difficulty	3 Severe difficulty	-3 Severe barrier	+3 Severe facilitator
4 Complete difficulty	4 Complete difficulty	-4 Complete barrier	+4 Complete facilitator
8 Not specified	8 Not specified	8 Not specified	8 Not specified
9 Not applicable	9 Not applicable	9 Not applicable	9 Not applicable

3.1.1.4 Application and benefits of the ICF-CY

The applications and benefits of the ICF-CY are not limited to classification tools, rather, they are also used as universal frameworks for research and clinical practice (12).

The ICF-CY is not a clinical assessment tool. However, it provides a means for the documentation of the level of functioning or delay in children with CP, not only for professionals but also for parents and caregivers (12). It also provides a basis for the design of individualised treatments through a structured goal-planning process (134). The ICF-CY is used in CP literature to describe the effects of various treatments (135-138).

In the research setting, selected ICF-CY categories and components have been used in comparison studies that assessed outcome measures and linked outcomes evaluations across studies. For example,

Wilson et al. (6) classified outcome measures that were used in the literature for lower limb orthopaedic surgery according to ICF-CY domains. Their in-depth analysis found that most studies focused on the 'body functions/structure' domain and only a few studies investigated other levels of the ICF-CY.

The scope of the surgical outcome domains and outcome measures that are used in the literature and their links to the ICF-CY framework are detailed in section 3.1.2.

3.1.1.5 Critique of the ICF-CY

Although there are significant advantages arising from the use of the ICF- CY in clinical care and research, its application remains difficult. One of the significant challenges is the large number of codes that are included in the system; institutions usually have insufficient resources and/or time to complete all codes of interest for a particular child, and this limits its practical use in the clinic (139, 140). Thus, reduced lists of ICF-CY categories have been recommended. These are referred to as 'core sets' (141). Core sets are comprised of carefully chosen ICF-CY categories that are intended to represent a minimum standard for clinical work and research for a specific health condition or disorder (further details are given in section 2.6 Core Outcome Set).

Another limitation of the framework is that it is difficult to apply the ICF-CY coding globally in different cultures. Different codes may have different meanings and implications in different care settings. For instance, the code for 'walking a short distance' can be interpreted differently due to cultural implications. In one society, 'walking within the home' may be interpreted as walking a short distance, while in another society, 'walking for about 2km' may be considered as short distance (132).

3.1.2 Assessment of lower limb orthopaedic surgery

Based on the ICF-CY, evaluation of primary surgical outcomes has historically focused on the improvement of impaired body functions and structure, such as joint range of motion, spasticity and gait parameters. These are measured through the use of relevant tools, for example: clinical examination, 3DGA, X-Ray, CT or MRI scan, or pain measures (6, 142). Ultimately, these outcomes are aligned with the objectives of orthopaedic surgical management in ambulant CP, which are to improve function, decrease discomfort, and prevent disabling structural changes (4, 143). A review of the literature on the evaluation of treatment outcomes in this field was conducted by Wilson et al. (6). A total of 229 articles were included. These outcome evaluation studies were usually limited to the ICF-CY domains of structure and

functions (91%). A small number (9%) analysed day-to-day activities and children's participation in daily life activities (6).

There is a common assumption that treatment aimed to improve one level in the ICF-CY framework, such as body structure and function (e.g., knee flexion contracture or crouched gait), should positively affect another level such as activity and participation (e.g., by enabling independent walking over longer distances than could be achieved before). Similarly, it is evident that interventions do not always have simple effects on a single dimension. In lower limb orthopaedic surgery, Gorton et al. (123) found that the tools that were used to measure outcomes were not sufficiently responsive to measure the effect of gait improvements on activity and participation, or that there were no such changes. This study was conducted through evaluation of the data from 75 ambulant children with CP who had undergone lower extremity surgery. Their post-operative changes were compared with those in a matched nonsurgical group. The study reported that surgery was effective in improving gait, but these improvements did not necessarily translate into notable changes in activity and participation (123).

There has been a move among clinicians treating CP toward optimisation of activity and participation (6, 142). Several studies have emphasised that the analysis of treatment outcomes must change in the future toward patient-reported outcomes (PROs) and the use of patient-reported outcome measures (PROMs) (3, 6). The change of treatment focus toward activity and participation aligns with the wishes of children's families, who are interested in what their child will be able to do after treatment, rather than changes in body structure and function. Vargus-Adams et al. (144) investigated the outcomes that were important to children and their families after surgery, and they showed that children and young people and their families were focused on what the child/young person would be able to do in everyday life (144).

The next section provides an overview of primary outcomes and outcome measures that are commonly reported in CP. These are discussed under the most relevant ICF-CY domain.

3.1.2.1 Body function and structures

Clinical examination

Clinical assessment, in particular for Range of Motion (RoM), strength and tone outcomes, is typically performed before and after lower limb surgical interventions in CP. Joint RoM measurements are widely employed by physiotherapists and orthopaedic surgeons to assess muscle shortening and joint contracture. It has been suggested that this should be a mandatory assessment to aid in the interpretation of kinematic data that are collected during gait analysis (145). Goniometry, or angle measurement, has been used to assess joint RoM; however, this measure has questionable reliability (145, 146).

Clinical examination is also performed postoperatively to assess the changes that occur in joint and muscle RoM as well as in muscle tone. In children with CP, the most common lower limb muscles that are affected by spasticity and require surgical intervention are the gastrocnemius-soleus (triceps surae), medial hamstrings, rectus femoris, hip adductors and psoas (147). Spasticity can be measured by the Modified Ashworth Scale (MAS) (148) or Modified Tardieu Scale (MTS) (149). A description of the grades of each scale is presented in Table 9. According to a study by Numanoğlu et al. (150), the MAS offers better clinical utility than MTS in terms of the time needed for its administration, but MTS has better intra-observer reliability.

Finally, muscle strength/power is measured pre-operatively, as part of the clinical assessment (to assist with surgical decision making) and postoperatively for comparison. Examination of the muscle strength around the ankle, knee and hip can significantly affect gait pattern and walking ability (147, 151). The Oxford Scale of muscle power has been used frequently to assess the weakness of lower limb muscles (i.e., isometric muscular force) (152). A description of the grades that are allocated is presented in Table 10.

Amirmudin et al. (153) reviewed published studies of the effects of SEMLS in children with CP and concluded that there was no postoperative change in spasticity or strength in the long-term, although there was improvement in joint RoM and spasticity at 6 and 12 months after surgery.

Table 9 Grade of Modified Ashworth Scale and Modified Tardieu Scale

Grade	Modified Ashworth Scale	Modified Tardieu Scale
0	No increase in muscle tone.	No resistance throughout the course of passive movement.
1	Slight increase in muscle tone, manifested by a catch and release or by minimal resistance at the end of the range of motion when the affected part(s) is moved in flexion or extension.	Slight resistance throughout the course of passive movement, with no clear catch at precise angle.
1+	Slight increase in muscle tone, manifested by a catch, followed by minimal resistance throughout the remainder (less than half) of the ROM.	N/A
2	More marked increase in muscle tone through most of the ROM but affected part(s) move easily.	Clear catch at precise angle, interrupting passive movement, followed by release.
3	Considerable increase in muscle tone, passive movement difficult.	Fatigable clonus (<10s when maintaining pressure) occurring at precise angle.
4	Affected part(s) rigid in flexion or extension.	Infatigable clonus (>10s when maintaining pressure) occurring at precise angle.

Table 10 Oxford Scale of Muscle Power

Grade	Manual Muscle Testing
0	No evidence of contractility (complete paralysis)
1	Slight contractility without any movement
2	The patient completes full or partial range of motion with gravity eliminated
3	Completes the available test range of motion against gravity, but tolerates no resistance
4	Movement against gravity plus some resistance
5	Movement against gravity with full resistance

Radiology

The assessment of skeletal malalignment through radiological examination is an important part of surgical decision-making for children with CP. Common examples of such misalignment are abnormal femoral anteversion and tibial torsion (154). These are identified clinically and on gait assessment but usually require radiographic confirmation. Information obtained through radiographs of the standing foot have been reported to be clinically relevant and have been used in children with CP (155). Hip subluxation and dislocation have been reported to be present in 11% of ambulatory children with CP (156). Thus, according to Cerebral Palsy Integrated Pathway Scotland (CPIPS) (157), children with GMFCS I and II are advised to undergo a pelvic radiograph at the ages of two, six and sixteen years old. Hip imaging for children with GMFCS level III is recommended yearly till eight years of age and should be repeated every two years thereafter (158).

Gait pathology/pattern

Gait deviations are present in children with CP, with patterns that are often complex because of the impaired neurological muscle control, the musculoskeletal deformities and the complex movement interactions between different muscles, joints and body segments. Gait assessment is valuable in defining surgical indications and determining the effects of the interventions on the child's gait characteristics. A systematic review by Amirmudin et al. (153) on the effects of SEMLS on children with CP found that 50% of the included studies measured gait parameters.

Several outcome measures are used to assess gait in children with CP. In orthopaedic surgery, 3DGA is frequently used as part of a preoperative assessment, in order to identify all gait deviations and plan treatment. Post-operative gait analysis is also used to assess the results of treatment. The 3DGA usually involves recording of kinematic and kinetic data of the joints in a laboratory setting and may also include dynamic electromyography (EMG) (159-161). Kinematic data describes limb segment and joint motion in linear and angular displacement, velocity and accelerations. The data are derived from an optoelectronic system, which is a sophisticated computerized video camera apparatus, that assesses the motion of each limb segment and joint in all three planes during gait. Kinetic data describe the measurement of joint moments and power, which are calculated using the GRF data obtained from the force plates. Inverse dynamics and simplified models of the musculoskeletal system are used for the calculations. Dynamic EMG provides information about the timing and duration of voluntary and involuntary muscle activity during gait through either surface or fine-wire electrodes (160, 161).

Several single numerical scores derived from 3DGA have been suggested to represent overall gait deviation from the norm. These include the Gillette Gait Index (GGI), Gait Deviation Index (GDI), and Gait Profile Score (GPS). Of these, the GGI was developed first; however, the difficulty in interpretating the data was considered to be a limitation in its use (162, 163). The GDI was developed to address this limitation. The GDI defined as a scaled distance between the 15 gait feature scores (163). It contained a smaller number of parameters than the GGI so that it was easier to interpret (163). The GDI has been used widely in studies of lower limb orthopaedic surgery in ambulant children with CP (164-168). The GPS provided information on segment and joint-specific deviations. It has been shown that the GDI and GPS offer improved sensitivity over GGI during the assessment of interventions (169).

A systematic review was conducted by McGinley et al. (110) to determine outcome measures that were used to assess the effectiveness of SEMLS in children with CP. They found that 3DGA was the most

commonly used outcome. A scoping review of outcome measures also found that 95% of studies in this field reported outcomes through the use of 3DGA (6).

Although 3DGA is an important and objective method of clinical assessment, it has limitations in that it produces a large amount of data and graphs; for this reason, despite its objectivity, its use can be complicated, and the data interpretation can be challenging and requires relevant expertise. Single number gait indices derived by gait kinematic and kinetic parameters, such as the GDI and GPS discussed above, do not reveal the source and nature of the abnormalities. The GPS can be presented together with a numerical or graphic representation of the multiple gait parameters on which it is based. This offers additional information on the source of the gait deviations (170). Further to this, gait analysis requires trained staff and appropriate equipment and space. The evaluation and interpretation of the results is time consuming (147). In this context, observational gait assessment is an important and useful alternative outcome measure for clinical use. It can be used to determine gait deviations and their changes. It is evident from a variety of studies that these scores have been used to assess pre- and postoperative gait.

The observational gait assessment measures include the Physician Rating Scale (PRS) (171), Observational Gait Scale (OGS) (172), Visual Gait Score (VGS), Edinburgh Visual Gait Scale (EVGS) (173), Observational Gait Analysis (OGA), and Visual Gait Assessment Scale (VGAS) (174, 175). The PRS and EVGS are the most commonly used observational gait assessment measures. The EVGS is considered the best option, as it has satisfactory reliability and concurrent validity and it provides information in all three planes for the foot, knee, hip, pelvis, and trunk in stance and swing (175).

Foot pressure

Foot stability and positioning at initial contact and throughout the gait cycle are aften assessed through measuring the distribution of pressure over the plantar surface of the foot (dynamic pedobarography). This is often collected simultaneously with the 3DGA. Pedobarography is useful in quantifying varus and valgus in particular for hindfoot deformities and in identifying areas of excessive pressure, which may give rise to problems. The pressure data is presented as a diagram on a coloured grid, which indicates pressure distribution over the plantar surface of the foot and its changes during the gait cycle (160). Although radiography is important in the preoperative assessment of the foot, pedobarography can offer additional information complementing radiography (176).

Energy consumption (expenditure)

Assessment of walking energy efficiency can be assessed by measuring (1) the amount of oxygen consumed and carbon dioxide expired, (2) indirectly, by measuring heart pulse when it has reached a steady state, or (3) the mechanical cost of work as determined by force plate data (160, 177). Thomas et al. (177) reported that energy consumption during walking is varied and depends on an individuals' weight, height and leg lengths. Measuring energy expenditure through oxygen cost before and after an intervention has been reported as a valid objective measure of change in gait energy efficiency for children with CP (178).

3.1.2.2 Activity and Participation

Assessment of functional level and motor development

Published CP studies have emphasised the importance of assessing a child's motor development, functional skills, and activity limitations, particularly when assessing the effect of an intervention. The Gross Motor Function Measure (GMFM) (179, 180) is a validated instrument that is frequently used to measure changes in gross motor function after an intervention. This tool has five different dimensions, including skills in supine/prone positions, sitting, crawling, standing up, and walking (181).

Activities of daily life

Everyday tasks that children perform in their school, home, and social environments are included in the activity and participation component of the ICF-CY framework. Outcomes of surgery that are relevant to activity and participation have been presented in previous studies using a variety of outcome measures (3, 182).

Several activities and participation outcome measures have been used in the context of lower limb orthopaedic surgery for CP. These include the Gillette FAQ (183), the FMS (184), the Pediatric Evaluation of Disability Inventory (PEDI) (185), the Pediatric Outcomes Data Collection Instrument (PODCI) (186), the Functional Independence Measure for Children (FIM) (187), the One-Minute Walk Test (1MWT) (188), the Six-Minute Walk Test (6-MWT) (189) and the Gait Outcome Assessment List (GOAL) (3). Table 11 summarises the features of these outcome measures.

The Pediatric Evaluation of Disability Inventory (PEDI) and Functional Independence Measure for Children (FIM) are well supported by the literature because they include items that measure activity in daily life. However, both were appropriate for only older children (4, 190).

Some of the above outcome measures are child or parent-reported or based on direct observation. Family members and caregivers are usually involved in responding to child-reported questionnaires, particularly in the case of children and young people with significant cognitive impairments or who have difficulty in communicating. Table 11 shows the clinical administration method for each of these measures.

3.1.2.3 Contextual factors

Environmental factors

Environmental factors are usually unchanged after an orthopaedic intervention and are generally not defined or included in the analysis of surgical outcomes. However, these factors are important when considering outcomes, particularly in order to offer children and families realistic expectations.

Personal factors

A child's personal factors may include age, gender, race, educational level and social background. These are not coded in the ICF-CY framework due to wide cultural variability (12). In clinical research, these factors are collected as part of participants' demographics if they are relevant to the research, however, these factors are important as they may affect a number of medical parameters (191).

3.1.2.4 Quality of Life

Several studies have reported outcomes related to quality of life (QoL) in children with CP (192-194). Viehweger et al. (142) supported the inclusion of QoL measures in the design of evaluation studies in paediatric orthopaedics. However, a review led by Wilson et al. (6) found that across 221 studies pertinent to lower limb orthopaedic surgery in CP between 1990 and 2011, only two studies had evaluated children's QoL status. This may be because researchers do not consider the potential impact of orthopaedic surgery on QoL.

Several QoL outcome measures have been developed and used in the literature, such as the CHQ (195), the CP-QoL survey for children (196), and the PedsQL (CP module) (197). Several studies recommended the CP-QoL child questionnaire as the preferred option because of its optimal psychometric properties and clinical utility (198-200).

Table 11 Features of Activity/Participation outcome measures

Outcome measures	Target population	Administration format	Numerical vs descriptive	Aspect of activity measured
Gross Motor Function Measure	0-12 years	Direct observation	Numerical measure	Gross motor activities: Lying and rolling, sitting, crawling and kneeling, standing, and walking, running and jumping
Gillette Functional Assessment Questionnaire	Unclear	Self-reported	Descriptive measure	Mobility
Functional Mobility Scale	6-18 years	Parent-reported	Descriptive measure	Mobility
Pediatric Evaluation of Disability Inventory	6 months- 8 years	Self or parent-reported	Numerical measure	Self-care, mobility, social function
Pediatric Outcomes Data Collection Instrument	0-18 years	Self or parent-reported	Numerical and descriptive measure	Physical function
Functional Independence Measure	7-18 years	Self-reported	Numerical measure	Self-care, mobility, cognition
1-Minute Walk Test/ 6-Minute Walk Test	All ages	Direct observation	Descriptive measure	Mobility
Gait Outcome Assessment List	9-18 years	Self or Parent-reported	Numerical measure	Activity of daily living and Independence, gait function and mobility, physical activities, game and recreation, use of braces and assistive devices

3.2 Children's expectations, preferences and priorities

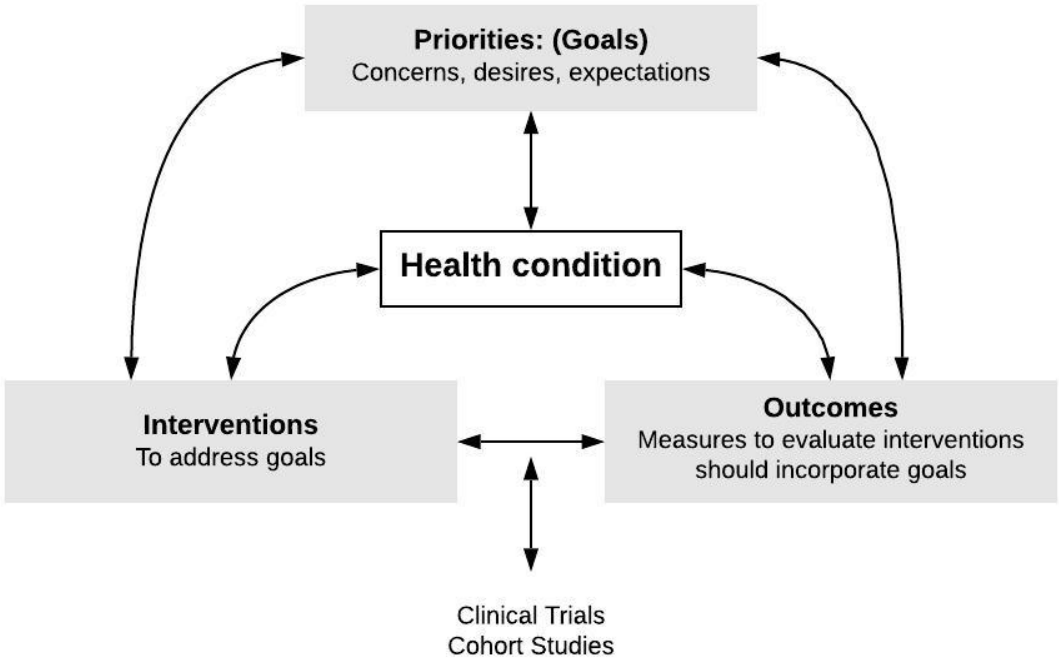
This section discusses the importance of assessing the preferences and priorities of children who are under consideration for surgery. This subject fits within the broader concept of healthcare quality following interventions. Understanding patient priorities during surgical decision making would help clinicians define the optimal post-operative outcome measures and avoid assessment of irrelevant surgical outcomes. Furthermore, this section discusses the challenges of measuring expectations, preferences, and priorities.

3.2.1 Children and families' important outcomes

Thomason et al. (3) emphasised the importance of understanding the priorities and expectations of the child with CP and their family when assessing the effectiveness of any intervention. There is increasing recognition that children should be consulted on their treatment goals and this information should be incorporated into treatment plans (201, 202). Narayanan (203) acknowledged that patients found outcomes to be most meaningful when they were aligned with their priorities. Similarly, Weiner et al. (204) stated that paying attention to patient needs and circumstances when planning care was associated with improved healthcare outcomes.

Narayanan drew up a priority framework for outcome assessment (Figure 8) (203). Health conditions of interest were placed at the centre of this framework. Priorities were influenced by an individual's concerns, desires, and expectations. Goals were a reflection of one's priorities and guided the decision-making process related to interventions. The effectiveness of these interventions was evaluated by outcome measures. For these outcome measures to be meaningful, they should, in turn, reflect the original goals. Moreover, the framework recognised that multiple stakeholders were involved in healthcare decision-making, and that although their perspectives might overlap, they may not be concordant.

Similarly, it is widely accepted that a family-centred approach to intervention improves motivation and outcomes (205, 206). This approach is based on the identification of needs and leads to the planning of interventions to meet the requirements of the individual's goals (207). This approach recognises that each family is unique; that the family is the constant in the child's life; and that they are the experts on the child's abilities and needs. (208).



Stakeholders' perspective

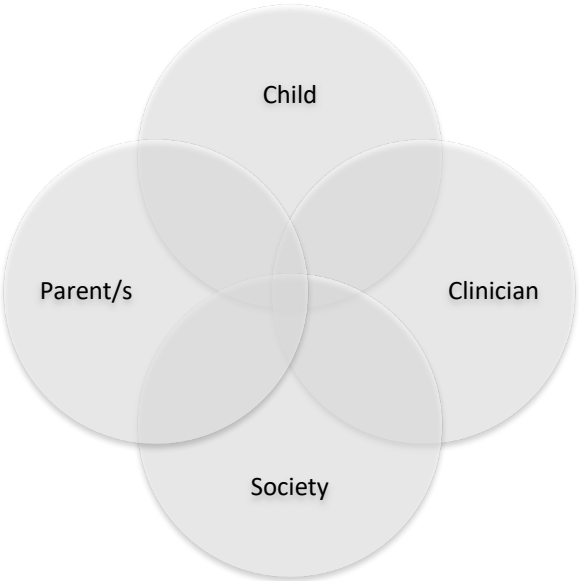


Figure 8 Priority Framework for Outcomes Assessment

Yet, little is known about the priorities and needs of children with CP and their families in the context of lower limb orthopaedic surgery. Few studies explored children's and parents' experiences following surgery. Capjon et al. (209) studied eight children (10-17 years old) and their parents' major concerns when a surgical procedure was planned. They identified that the main priorities of both children and

parents were (1) the management of the deterioration of physical capacity, which could otherwise result in pain and fatigue, (2) being socially accepted and (3) improved life for the child. Lehtonen et al. (210) interviewed ten children and young adults, (between ten and 22 years of age) with CP, to explore their experiences five years after they had undergone SEMLS. The researchers found that their motivation for undergoing the surgery was maintenance of their mobility levels and physical activity; retention of future walking function; being independent; and participation in a real-life environment. Similarly, Capjon et al. (211) interviewed eight children and their parents to explore their experiences regarding rehabilitation following SEMLS. They found that the rehabilitation and its level of success had a great impact on the child's physical functioning and social participation, which were linked with their satisfaction levels. However, the parents stated that they lacked information and effective interconnection between related health services. Although these studies were based on a small sample size, qualitative research depended on saturation of the data and the identification of themes, which were acknowledged in these studies.

3.2.2 Challenges

Two challenges contribute to the difficulty in assessing preferences and priorities. Firstly, each child and family have different needs (212). Further to that, expectations and priorities change over time as life events that are associated with the child's development and symptoms change (213). Secondly, parents usually make decisions on behalf of their children and in accordance with their own and their child's experiences (212, 214).

To address these challenges, the goals of treatment should be realistic and should enhance the functional ability of the child. The child and family must be encouraged to take a central role in the decision-making process. This practice can identify and address any unrealistic expectations before the operation and may be a key factor in the overall success of and satisfaction following the surgical intervention.

3.3 Limitations in the Evidence-Base for Orthopaedic Surgery in CP

This section provides a review of the primary limitations in the evidence base that supports the use of orthopaedic surgery in children with CP. The literature review presented below will demonstrate heterogeneity in outcome reporting, variation in the selection of outcome measures, choice of outcomes that are irrelevant to children and parents, selective reporting bias and an overall poor quality of evidence.

3.3.1 Heterogeneity of reported outcomes

There is no guidance available regarding the outcomes which researchers and clinicians should report following orthopaedic surgical interventions for children with CP. A variety of outcomes have been used and inconsistencies in outcome reporting have been observed. Williamson et al. (215) emphasised that "the problem of the inconsistency of reporting outcomes is often highlighted when completing systematic reviews and meta-analysis". Systematic reviews and meta-analyses are used to summarise the published evidence and assess the impact of an intervention by summing and analysing the published data in their totality. However, if published studies do not assess the same outcomes, such conclusions cannot be drawn. Concerns over the heterogeneous nature of outcome reporting and its impact have been reported in several systematic reviews of primary studies that considered the effects of lower limb orthopaedic surgical interventions on children with CP (5, 110, 153, 216).

A systematic review of 31 studies was performed by McGinley et al. (110). The review explored the quality of the evidence for SEMLS. The authors found that the majority of studies that were included in the review reported outcomes under one ICF domain only, and mostly fell in the body function and structure field (e.g., gait-related outcomes). There was less evidence available on outcomes relevant to activity and participation or QoL. Although the reported SEMLS outcomes were synthesised into conclusions, the authors stated that "a meta-analysis of the results was not considered appropriate given the heterogeneity of study … reported outcome domains" (110).

Lamberts et al. (5) systematically reviewed 24 studies on the effects of SEMLS on children's gait parameters. The review reported improvement in gait parameters after the SEMLS intervention. However, this review was based on only one surgical outcome domain (gait parameters). The authors suggested that it was not possible to conduct a meta-analysis and to draw clear, overall conclusions due

to the heterogeneity in the gait parameter outcomes that were considered in the included studies; 19 gait parameters were reported within Lamberts et al.'s systematic review.

In a review by Edwards et al. (216), the authors aimed at identifying potential predictors of and factors that affected outcomes after SEMLS in children with CP. They defined gender, topographical distribution, and socio-economic status as predictive factors. Although this review was based on only seven primary studies, the authors reported that it was not possible to compare the studies due to the heterogeneity of the measured outcomes. This was also stated by Amirmudin et al. (153) who reviewed 74 studies to summarise the effects of SEMLS and found moderate heterogeneity between them.

Similarly, the impact of heterogeneity between studies that have been included in systematic reviews has been confirmed in the area of rehabilitation following surgery. Bommel et al. (108) reviewed six studies on physiotherapy rehabilitation following SEMLS. The authors noted that there were incomplete descriptions of the interventions and the methods they used. There was a variety of interventions with different goals, frequency, duration and intensity in all the studies. The authors concluded that "because of the variation and the lack of description of the (physiotherapy), it is difficult to draw straightforward conclusions related to the (physiotherapy)".

Of these outcomes, most were found to relate to the body function domain of the ICF-CY. On this basis, it could be concluded that impairment level outcomes, rather than activity or participation outcomes, form the primary aim of lower limb orthopaedic surgical interventions. However, this does not address the recent shift in the aim of surgical interventions, i.e., the focus on the child's activity and participation level. There is a need for consensus on outcomes that are most appropriate from the child/family and the clinician's point of view to assess results after lower limb orthopaedic surgery for CP.

3.3.2 Heterogeneity of outcome measures

Numerous outcome measures are used following lower limb orthopaedic surgical interventions. A review of the outcome measures that are used in CP studies in this field (6) demonstrated the variability of these measures. In this review of 229 studies, 32 different outcome measures were used. In addition, a range of informal, individualised and insufficiently described assessments were used to measure the effects of interventions. The authors noted the wide range of outcome measures and highlighted the need for improvements in the quality of future studies; unbiased reporting and the use of standardised outcome measures were recommended (6).

McGinley et al. (181) encountered similar issues in their systematic review, which examined the quality of evidence that supported SEMLS for the CP population. The authors also noted the use of inappropriate outcome measures. For example, the GMFCS level was used incorrectly as the primary and only outcome measure in two studies (217, 218). Further, in a recent review by Amirmudin et al. (153), the authors reported that heterogeneity and inconsistency were identified in the measures that were used to assess each SEMLS outcome domain. Wilson et al. (6) acknowledged this problem and stated "few outcome measures used consistently across the literature aimed to measure similar outcomes".

The variability that is evident in the outcome measures used in CP orthopaedic surgery research may be attributable to an increasing number of available measures and the absence of synthesised information regarding their psychometric properties and content. For example, Wilson et al. (6) identified 14 outcome measures that had no published psychometric properties, yet they had been used in 68 studies.

Zanudin et al. (219) systematically reviewed the psychometric properties of 18 outcome measures that were used to assess gait quality and walking performance in young people with CP. Psychometric properties including *reliability*, which is defined as the ability to reproduce a consistent result in time and space, or from different observers; *validity*, which is referred to the fact that a tool measures exactly what it is proposed to measure, were evaluated using a modified checklist based on the COnsensus-based Standard for the selection of health status Measurement Instruments (COSMIN) (220). Although the identified outcome measures showed moderate to strong evidence for one of their psychometric properties, none of the outcome measures had a strong level of evidence for all psychometric properties.

Therefore, increased uniformity in the outcome measures used in research and strong evidence of their psychometric properties would be required to facilitate the combination and comparison of research results and the meta-analysis of research outcomes.

3.3.3 Unmet expectations, irrelevant outcomes

Research has shown that, in many cases, interventions that involve rehabilitation programmes for children with CP are not aligned with the expectations, goals and needs of the children and their families (139, 221-223). High-quality evidence for the effectiveness of healthcare treatments should derive from high-quality studies that assess outcome measures relevant to patients (224).

Currently, surgical outcomes such as gross motor and gait functions are assessed through the application of different clinician-administered measures. Although these assessments provide a wealth of objective information about a child's motor function, they generate little information about the child's or parents' views of functioning, and do not attempt to understand the priorities or expectations of the child or family members (3). For example, surgical interventions have been guided by gait analysis for over 20 years. However, there is little information as to how reliably post-operative improvement of gait analysis data can be used as a primary outcome measure to reflect changes in a child's daily walking activity, which is usually considered to be one of the child's and family's priorities (3, 225).

Patient-Reported Outcome Measures (PROMs) are considered to offer the best method of evaluation, because they measure activity and participation. The PROMs enable children and families to assess elements of their health and related function. This is essential because the child and family members have first-hand knowledge of the child's daily functioning in all contexts, so they are the most qualified to report on this (226). Narayanan (203) states: "To the extent that the patient's perspective is recognised to be pre-eminent in making judgments about effectiveness, the use of PRO measures is now considered the standard when evaluating the effectiveness of interventions."

This is, however, rarely the case in published studies of lower limb orthopaedic surgery in CP. Clinician-administered outcomes are often used in clinical studies, including clinical examinations, 3DGA and radiological investigations (6, 227). A recent review highlighted the lack of PROMs used in the field, as only 20% of the outcome measures used were referred to be PROMs (227).

PROMs allow direct patient assessment. However, in young children or those with cognitive problems, the assessment is often completed by a family member or caregivers by proxy (142). However, a few studies have shown that there can be lack of agreement between different responses. For instance, Radsel et al. (228) conducted a cross-sectional study to assess the Health Related Quality of Life (HRQoL) of children with CP (aged 6-16 years old). They found differences in questionnaire scoring between caregivers and children. Proxy and self-reported versions of the questionnaires (DISABKIDS generic) (229) were used. Among 91 caregivers and 48 children who filled out the questionnaires, the authors found that children self-reported higher scores than their caregivers (mean score 75.6, standard deviation (SD) 15.9 versus mean 72.3, SD 17.9). Adolescents scored lower than children in all DISABKIDS domains (mean score 69.4, SD 19.4 versus mean 80.8, SD 10.0). Similar differences between children's and caregiver's scores have also been reported by other studies (230, 231).

It has been suggested that children with CP, their families and their clinicians seek different goals from health interventions. Vargus-Adams (144) who assessed what outcomes were important to children (n= 17), families (n=19) and medical professionals (n= 39). This showed that children and families were more focused on how any suggested interventions would change what the child's ability in everyday life. The author concluded that "with a greater understanding of the important domains for assessment in childhood CP, better means of measuring change may be developed" (144).

There has been a long-standing assumption that changing the impairment caused by CP would lead to better function and engagement in life activities (65). In a study conducted by Vos et al. (232), it was shown that joint range of motion and spasticity had no or little effect on gross motor function. These impairment level outcome measures are not viewed as important to children and their families. Thompson et al. (182) suggested that the timing of when treatment is offered also impacts on goal setting. Goals that were set before surgery were likely to have an impairment-based focus, such as correction of musculoskeletal deformities. In contrast, goals that were set during post-operative rehabilitation were likely to be functional and to focus on activity and participation as well as HQoL.

Understanding and assessing patient expectations from surgical interventions are also central to the decision-making process. When guiding patients towards the right decision it is important to discuss any unrealistic expectations from surgery. It has been shown that discussing patients' expectations pre-operatively, and offering advice on lifestyle adjustments to meet postoperative needs, enhances patient satisfaction and improves outcomes (233-235).

For the reasons discussed above, it is important that clinicians and researchers clearly identify and prioritise outcomes that are important to patients. Research that purposefully examines the desired surgical outcomes of all relevant stakeholders is required to inform and guide research and clinical practice and to enable patients to make informed decisions about treatment.

3.3.4 Selective reporting bias

Systematic reviews and meta-analyses offer the best available evidence in an area of clinical practice. Kirkham et al. (236) defined the purpose of a systematic review of healthcare as follows: "Systematic reviews of clinical trials aim to include all relevant primary studies conducted on a particular topic and to provide an unbiased summary of their results, producing the best evidence about the benefits and

harms of interventions" (236). It is acknowledged that the prevention of surgical harm is essential in the provision of high-quality clinical care, the reduction of harm to stakeholders, the increase of function, and the containment of healthcare costs (237-239). It is also evident that increased understanding of adverse surgical events guides the shared decision-making process with patients and families (237, 240).

Several studies emphasised the use of clear documentation and the collection of data regarding the occurrence of adverse events related to surgical interventions (237, 240-242). However, a series of reviews in lower limb orthopaedic surgery in children with CP have shown that adverse event outcomes are under-reported.

A literature review on the effects of SEMLS in children with CP (153) found that adverse events were only reported in 17/74 studies (23%). The authors reflected on the impact of this problem in their systematic review: "We can therefore not make conclusions regarding the safety of MLS in this review" (153). The review also found considerable variation in the way adverse events were reported. The complications identified by this review included intraoperative haemorrhage, poor postoperative pain management, hardware removal due to pain, superficial wound infection, nerve palsy, avascular necrosis of the femoral head, long-standing reflex sympathetic dystrophy, and development of genu recurvatum (153).

A standardised classification system for reporting the severity of adverse events related to orthopaedic surgery has been suggested: the Clavien and Dindo method. The Clavien and Dindo method is a five-grade classification system for adverse surgical events. Zhou et al. (242) evaluated the reliability of the modified Clavien and Dindo classification system (243) following lower limb orthopaedic surgery in children with CP, and they reported good reliability.

Reporting surgical adverse events is an important issue that must be addressed to improve transparency in the identification and reporting of harm (adverse events) during surgical interventions.

3.3.5 Quality of the evidence

To date, the assessment of surgical interventions for children with CP has been primarily based on cohort studies. A review of the literature revealed several systematic reviews, which highlighted concerns over the quality of the current evidence.

The majority of studies in this field are retrospective and lack controls for comparison (6, 119, 244, 245). In 2014, more than 80% of research on lower limb orthopaedic surgery used retrospective design (6). Therefore, the existing evidence does not support the development of robust management guidelines, nor can it inform the design of RCTs to develop such guidelines.

RCTs to assess paediatric surgical conditions and their management present particular challenges. This is due to several factors, including ethical issues around assent, consent, investigator bias and the lack of agreement on which patient factors affect outcomes (246-248). Children with CP represent a heterogeneous population, with variable clinical characteristics. Therefore, recruiting homogeneous patient cohorts for adequately powered trials is challenging. The need for long-term follow-up in paediatric conditions and the effects of growth on outcomes also add to the difficulties of performing RCTs (249-252).

In the case of SEMLS, the size of published surgical studies is small. There are very few studies that are large enough to allow conclusions to be drawn on the benefits and harms of surgical interventions. Demographic information and heterogeneity of the clinical condition within the primary study populations, surgical technique and post-surgery rehabilitation are inadequately defined and described.

In 2012, McGinley et al (110) conducted a systematic review of the literature published between 1985 and 2010 on the outcomes of SEMLS. The review aimed to determine the quality of studies conducted on the outcomes of SEMLS. A total of 31 studies met the inclusion criteria and were assessed for the review. The quality of the included studies was assessed using the Methodological Index for Non-Randomised Studies (MINORS). Although there was an overall improvement over the years in the quality of the studies that were conducted, some recently published studies scored poorly and were of very low quality.

Concerns over the published literature included the small sample sizes, the lack of details regarding rehabilitation, and inadequate recording of the type of procedures and any complications. The advantages of this review were that a large number of articles were included and strict inclusion criteria were used. Furthermore, the two authors undertook the reviews separately and then discussed the findings. The review was then compiled after consultation with a more experienced professional in the field, which helped to decrease bias. The final conclusion was that further research in the field should be conducted as RCTs. These would offer a higher level of evidence and add value to the current body

of knowledge. The authors also determined that prospective studies were more beneficial than retrospective studies as they decreased bias (110).

Bischoff (96) compiled a systematic review that focused on the outcomes of the research as well as the quality of the research conducted. The review concluded that the follow-up period in the existing studies was too short and that the type of surgical procedures used differed. Furthermore, the author noted that there was no comparison between single-level and multi-level surgeries and that no RCTs on SEMLS had been conducted. Taking these factors into consideration, the author concluded that the current level of evidence to support SEMLS in children with CP was low. Bischoff also concluded that long-term follow-up was essential and that there was no sufficient literature to validate the advantage of SEMLS over single-level surgeries (96).

Narayanan (66) undertook a review of the literature in 2012, and also stated that the previous comparative studies involved small sample sizes and only short-term follow-up. Furthermore, there was no evidence that any improved function was due to the SEMLS or to the intensive physiotherapy that the child received after surgery. The outcome measures that were used showed significant change and included perceived outcomes. Little change was noted in functional outcome measures.

It is evident from the above reviews that further research in this area is required. There is a need for good-quality, high-level evidence and long follow-up to validate the specific procedures that are used to correct specific deformities (66, 110, 182). This is not only because of the lack of RCTs but also because few comparative studies have been conducted (66, 96). Researchers have called for more prospective, well-designed studies to be conducted rather than retrospective ones. The choice of surgical outcomes and adequate length of follow-up period have not been agreed. The outcome measures to be used should consider the actual function (as opposed to perceived function) at all levels of the ICF- CY (66, 96, 110).

3.4 Core Outcome Set

This section explains the role of a COS and its potential to overcome several limitations associated with the current evidence in this field, as described in the previous section. Several areas are discussed in this section, including the importance of the COS concepts, critique and the different initiatives that have previously aimed to develop COSs. Finally, any existing COS used in CP will be explored. This will define the knowledge gap in this field of lower limb orthopaedic surgery, which this thesis aims to address.

3.4.1 Overview

A COS is proposed as a method to reduce heterogeneity in reported outcomes used by researchers and clinicians and use patient-relevant outcomes. It also aims to reduce the risk of reporting bias and enable meta-analyses (9, 253, 254). A COS is a group of agreed, standardised outcomes that should, at a minimum, be reported in every study that is within the scope of the COS. This core set should be consensually agreed upon by all relevant stakeholders (e.g. healthcare professionals, researchers, policy-makers, funders, patients and the public) (10, 253).

The use of a COS does not prevent the use of additional outcomes, but rather represents the minimum set of outcomes that should be collected and reported. The primary outcome of a trial would most often be one of the COS outcomes. However, the primary outcome(s) in a trial should be chosen individually (10, 215, 253). The selection of primary and secondary outcomes should be specific to the research question of interest (215).

The development of a COS requires the reaching of a consensus through a stepwise approach: firstly on 'what' to measure (i.e. the core outcome domains that should be measured), and secondly on 'how' and 'when' to measure (i.e. the core outcome measurement instruments that should be used to measure the core domains) (215, 255).

3.4.2 Importance of COS development

There are many reported benefits from the use of COSs. Primarily, core sets have been identified as a means of reducing the number of missing outcome data in effectiveness trials and of improving the quality of systematic reviews. The standardisation of outcomes would also facilitate the comparison and combination of research data across studies while enabling researchers to explore study specific outcomes (215, 256). In conditions in which COSs have been in use for a significant period, they have

been shown to reduce heterogeneity in the reported outcomes (257). Kirkham et al. (258) investigated missing patient data in Cochrane systematic reviews and surveyed the coordinating editors of Cochrane Review Groups (CRGs) regarding the standardisation of outcomes. It was found that 73% of the coordinating editors suggested that a COS for effectiveness trials should be used routinely in a summary of findings tables.

An additional benefit of COS development is the incorporation of the perspectives of a wide range of stakeholders, with an emphasis on patient involvement. This process enables increased patient-centred practice in research, which leads to increased relevance and meaningfulness of research. For instance, the organisers of the outcome measures in a rheumatology (OMERACT) initiative, developed a COS with patient involvement in the consensus process. This led to the identification of fatigue as an important outcome that had not been previously considered (259).

The use of COSs in clinical trials is now a prerequisite for funding. For example, the National Institute for Health and Care Research (NIHR), the largest funder of healthcare research in the UK, encourages the use of COS. In their grant application process, they request that COSs be used if they exist in the area of healthcare to which the trial applies (260). Thus, COSs are becoming more frequently adopted by researchers as the starting point for the identification of trial outcomes (261). Guidelines such as those for Standard Protocol Items: Recommendations for Interventional Trials (SPIRIT), CONsolidated Standards of Reporting Trials (CONSORT) and an Instrument for reporting Planned Endpoints in Clinical Trials (InsPECT) for trial protocols have recognised the importance of COSs and of improving the scientific approach to choosing outcomes in clinical trials (262, 263).

3.4.3 Core outcome set critique

While COSs are valuable, they undoubtedly have limitations and challenges. First, no COS can capture every aspect or outcome that is relevant to every individual. Second, as the body of research increases and different outcomes are found to be important, the COS can be modified to ensure that the most relevant aspects of health are taken into account. For example, if studies to create a COS repeatedly bring up a concept (outcome) that is not in the published ICF-CY core set (264), yet it is found to be important to stakeholders, then it may be necessary to add this outcome to an updated version of the ICF-CY core set.

Kirkham et al. (258) investigated the opinions of the Cochrane Review coordinating editors regarding perceived challenges that were associated with the standardisation of outcomes in their particular Cochrane Review Groups. The reported challenges were primarily related to the process of developing COSs, the scope of available COSs and the uptake among researchers. To mitigate these challenges, they suggested regular refinement of available COSs. The development of specific COSs may be necessary for specific populations and interventions (for example, pharmacological versus non-pharmacological treatments).

3.4.4 Core Outcome Set Initiatives

Various groups have worked on the development of COSs. Some were focused on a specific intervention or health condition (Table 12).

Table 12 Core Outcome Set Initiatives

Initiatives	Purpose	Reference
Initiative on Methods, Measurement, and Pain Assessment in Clinical Trials (IMMPACT)	Develop consensus reviews and recommendations for the handling of pain in adults and children	(265)
Harmonising Outcome Measurement for Eczema (HOME) initiative	Involve an international group that works to develop a COS for eczema trials	(266)
CoRe Outcomes in Women's and Newborn (CROWN) initiative	Harmonise outcome reporting in women's health research	(267)
Outcome Measures in Rheumatoid Arthritis Clinical Trials (OMERACT)	Improve outcome measurement in rheumatology through the development and implementation of COSs	(255, 268)
International Consortium for Health Outcomes Measurement (ICHOM)	Develop agreed sets of outcomes to be used in clinical trials and practice that reflect what matters most to patients	(269)

3.4.4.1 The COMET Initiative

The COMET Initiative (www.comet-initiative.org) was founded in 2010 to facilitate the development and application of COSs in all areas of healthcare (10). The specific objectives of the COMET are to: (1) raise awareness of current problems with outcomes in clinical trials; (2) encourage COS development and uptake; (3) promote patient and public involvement (PPI) in COS development; (4) provide resources to facilitate these aims; (5) avoid unnecessary duplication of effort; and (6) encourage evidence-based COS development (10, 270).

The COMET initiative is focused on what should be measured and reported in all trials of a particular health condition, followed by the determination of how these outcomes should be measured. The

COMET initiative has developed methodological guidelines for COS development (10), with the production of a list of protocol items for COS studies (Core Outcome Set-STAndardised Protocol, or COS-STAP) (271). Other recent developments include the quality assessment checklist of COS studies (Core Outcome Set-STAndards for Development, or COS-STAD), a clear and transparent reporting guideline for COS development studies (Core Outcomes Set-STAndards for Reporting, or COS-STAR) and guidelines for the selection of outcome measurement instruments for COSs (Core Outcome Measurement Instrument Selection project, or COMIS) (272-274).

The COMET website includes a database, which is a source of studies relevant to the development of COSs. The database currently contains over 800 references for planned, ongoing, and completed work on COSs of different health conditions. Usage of the website and database has increased significantly over time, reflecting the growing interest in this field (260).

3.4.5 Existing COS for Cerebral Palsy

The COMET database and ICHOM were searched to identify existing knowledge and COS research in the field of CP. No COS study was registered in ICHOM, while the COMET search identified six COSs (7, 275-279). In addition to the COMET findings, a scoping search of the literature found a further three published COSs (280, 281).

Four of the identified COSs (Table 13) were excluded because they did not follow the COMET principle of COS in considering what is important to all stakeholders and in measuring outcomes after interventions.

Table 13 Excluded core outcomes sets

Authors (year)	COS focus	Reference
McPhee et al.	Multi-morbidity risk assessment and measurement of outcomes in individual with CP	(277)
Benner et al.	Cardiometabolic disease in adults with CP	(275)
Eliasson et al.	Identified which research question is important and should be prioritised in the use of Constrained-Induced Movement Therapy (CIMT) in children with CP	(276)
Van Brussel et al.	- Reviewed all studies of childhood conditions including CP. - The authors provided a core set to develop a framework for a disease-specific cardiopulmonary training programme and recommended clinical measures to be used in research	(278)

In 2009, Vargus-Adams et al. (7) emphasised the importance of developing consensus on the important outcomes that should be measured after healthcare interventions in CP. They used a Delphi survey among children and young people with CP, parents and healthcare professionals. A consensus was reached on eight different domains, which were: impairment, general health, gross motor skills, self-care/fine motor skills, speech/communication, integration/participation, quality of life, and caregiver issues. In addition, medical professionals suggested a choice between 10 to 27 outcome measures that could be used to assess each domain; the most common were the GMFM and CP-QoL questionnaires. However, the study participants presented with a diverse range of impairments, severities of condition and a variety of healthcare disciplines. These broad results may limit adaptation within specific clinical research.

Maenpaa et al. (279) conducted a consensus study on outcome measures that defined functional ability in children with CP in physiotherapy and occupational therapy settings. A consensus was agreed through reviewing the evidence and expert opinion on using the following measurement tools: (I) in physiotherapy: muscle function, Goal Attainment Scale, GMFCS, GMFM-66, 6MWT, Physiological Cost Index, Timed Up and Go (TUG), and a Movement Assessment Battery for Children (Movement-ABC). (II) In occupational therapy: the Munchener Functionellen Entwicklungsdiagnostik (MFED), Assisting Hand Assessment, Canadian Occupational Performance Measure (COPM), Bruininks-Oseretsky Test of Motor Proficiency (BOT-2), The Beery-Buktenica Developmental Test of Visual-Motor Integration (Beery-VMI), Quality of Upper Extremity Skills Test (QUEST), and Miller Function and Participation Scales (known as M-FUN). Unfortunately, the findings of this study cannot be generalised as the consensus was based on the Finnish population only and involved a limited range of clinical expertise.

Schiariti et al., conducted comprehensive studies between 2013 and 2017 to develop a COS within the ICF-CY framework for children with CP who were younger than 18 years old (141, 264, 281-285). The initial phases of this study involved a survey questionnaire and interviews with children and their parents that were aimed at developing preliminary core outcomes important to all stakeholders (283, 284). However, children with different levels of severity were included, which may have influenced the results.

The final phase of the above study included a consensus meeting with researchers and healthcare professionals. Consensus was reached on one comprehensive and four brief core sets. The comprehensive set involved 135 outcomes; the common brief set included a minimum set of outcomes on 25 categories that fell within the ICF-CY framework (body functions (n=8), activities and participation

(n=8), environmental factors (n=8) and body structures (n=1)) (264). Although there were representatives from five different countries, the vast majority of participants were from the USA. Therefore, the views of under-represented populations were not included.

A systematic review was also undertaken by Schiariti et al. (285) to identify outcome measures aligned with the core set of outcomes. The review identified 83 outcome measures, of which 68 had been tested for their reliability and validity. The authors demonstrated that 25 valid and reliable measures were aligned with the defined core set. Although these measures showed satisfactory reliability and validity, most of them had been developed and tested in different conditions and not in CP.

Since paediatric neurodisability disorders are among the most burdensome health conditions and CP is recognised as the most common cause of childhood disability worldwide (1, 2), a search for a COS in neurodisability is important. A series of studies have aimed to develop a COS for this population (286-289). The authors of these studies presented core health domains that were important from the perspectives of all stakeholders (i.e., parents, children, and health professionals) and the accompanying measurement instruments. The domains included communication, emotional wellbeing, pain, ability to sleep, mobility, self-care, independence, mental health, community and social life, behaviour, toileting and safety.

This study gathered the views of a wide range of young people and parents from diverse backgrounds and areas around the UK. However, the individual subgroup population were different and there was a broad range of participant characteristics. This would have affected the identified outcomes, since key outcomes differ between different levels and types of neurodisability.

3.4.6 The need for a COS in CP/lower limb orthopaedic surgery

The diversity of opinions on the management of a condition may present a barrier to the development of a single COS within that field, and this may indeed be the case in CP. It has been suggested that further refinement through the development of multiple COSs for CP would be necessary to develop for specific interventions (258). Lower limb orthopaedic surgery is one of these interventions, as many children with CP undergo surgery to address lower limb secondary muscle contractures and bone deformities to improve or maintain their mobility.

This surgery is complex and resource-intensive, requiring a significant investment of time, effort and resources by the patient, family and healthcare system, and requires extensive rehabilitation. Currently, no consensus-based, standardised and agreed outcome domains and measures exist for international use. It is important, therefore, to develop a COS in this field. This will produce uniform and comparable surgical outcome domains and measures by identifying the most important outcomes for healthcare professionals, children and parents and by meeting the goals of all stakeholders involved in this intervention.

3.5 Summary and conclusions

A review of the literature confirms that there is heterogeneity in the outcomes measured in research into CP lower limb orthopaedic surgery. Consensus on important outcomes in this field is required to ensure that research is relevant. It is proposed that core outcome domains and outcome measures for CP orthopaedic surgery should be standardised through the development of a COS. This would reduce the current heterogeneity in reported outcomes and improve the quality of evidence. It would also facilitate the comparison and combination of research outcomes through meta-analyses and systematic reviews. The incorporation of core outcomes into research studies should also encourage greater transparency in research reporting by reducing selective reporting bias. The project methodologies that are used throughout this thesis are based on the recommendations from the COMET initiative (10), but adapted to the needs of this specific population and the study objectives.

Chapter 4 Research Aims, Objectives and Rationale

4.1 Research aims and objectives

The aims and the specific objectives of the research project described in this thesis are as follows:

1: to develop an internationally endorsed, COS for lower limb orthopaedic surgery for ambulant children with CP that has relevance to all key stakeholders:

A. to understand the experiences of lower limb orthopaedic surgery among children with CP and their caregivers,
B. to develop a comprehensive list of measurable outcomes of surgical interventions informed by literature and qualitative interviews,
C. to identify the outcomes that are most important to all stakeholder groups, and
D. to develop consensus from all stakeholder groups in order to define the final list that would constitute a COS.

2: to develop an international consensus on the selection of outcome measurement instruments for outcomes that would be included in the COS:

A. to identify outcome measures that have been used to evaluate lower limb orthopaedic surgery in CP,
B. to assess the literature evidence on the psychometric performance and feasibility of these measures in children with ambulant CP, and
C. to develop consensus from healthcare professionals in order to define the final list of measures that would constitute the core outcome measures set.

4.2 Research Rationale

As CP is the most common cause of physical disability in childhood, it is one of the most studied conditions in childhood disability. International experts in the field of paediatric disability have focused on various aspects of CP. With regards to the musculoskeletal manifestations of CP, surveillance registries have been established to identify and treat problems early (for instance, hip dislocation surveillance (158, 290)). Despite these initiatives, what remains missing in the field of orthopaedic surgery in CP is the application of a 'standard language' to describe outcomes and outcome measures after surgery in this population.

In 2016, the USA National Institute of Health (NIH) organised a symposium on CP that involved stakeholders from academic, clinical, regulatory, and advocacy backgrounds (291). The main goal of the symposium was to discuss the major challenges and requirements to move forward with clinical research in CP. They discussed the challenges around health evaluation outcomes and stated: "There is often a divergence between the outcomes reported in clinical and research contexts and the perceived relevance for patients and observers/caretakers." Hence, they emphasised that, in future CP studies, health evaluation outcomes and outcome measures should be relevant to the children and their caregivers.

The James Lind Alliance (JLA) brings patients, parents and clinicians together in priority-setting partnerships (PSPs) to identify and prioritise the most important unanswered questions or evidence uncertainties. In 2019, Vella-Baldacchino et al. (292) conducted a JLA project in the UK aimed at the identification and prioritisation of research questions regarding surgical management of children with conditions that affected the lower limbs. Children, parents and healthcare professionals gathered in face-to-face prioritisation workshops. An optimal way to measure outcomes post- lower limb surgical intervention was agreed to be the most important research priority. The development of COSs would address the issues raised by stakeholders who were involved in both the NIH symposium and the JLA studies.

Another rationale for this project is the necessity to define the outcomes that are important to those affected by the condition and are candidates for lower limb orthopaedic surgery. These outcomes are important in defining the success of treatment. Thompson et al. (293) stated: "If we can assess and understand family priorities and expectations, we may be able to align the aims of clinicians with those

of families and improve outcomes of interventions for the child and satisfaction" (293). Knowledge of patient preferences is essential to improve shared (patient-doctor) decision-making, which is believed to enhance the quality of healthcare. The identification of outcomes that are meaningful to children with CP and their caregivers, and the development of instruments to measure these outcomes are the aims of the proposed COS development.

Identification of such outcomes, which are relevant to all stakeholders, will inform the development of large-scale observational studies. These studies will in turn address the issues that currently prevent the conduct of RCTs as described in 3.3.5, including specification of the condition of participants selection, sample size and availability of control group. Eventually, enough high-quality RCTs will have been conducted to allow their synthesis in systematic reviews and meta-analyses, upon which robust management guidelines can be based. This type of research in lower limb orthopaedic surgery is lacking due to the heterogeneity in the CP population. This represents the first step in a paradigm shift toward evidence-based patient care in CP surgery. Thus, the proposed COS project has the potential to impact significantly on clinical practice because it will be the first crucial step to facilitate high-quality RCTs in the field. The systematic use of the outcomes that are relevant to all stakeholders and the use of similar measurement tools across all studies will facilitate comparability of results, which will ultimately improve therapeutic interventions. Therefore, there is a need for a COS for this population and intervention.

Amirmudian et al. (153) found that few studies in this field have assessed surgical outcomes according to ICF-CY domains, despite the recognised advantages of the framework in health evaluation and health-based research (12, 134-138). The lack of ICF-CY use may be explained by the extensive scope of the framework, which contains more than 1600 codes. Support from professionals nationally and internationally was required to make the ICF-CY as user-friendly and meaningful as possible. Schiariti et al. made efforts to facilitate the acceptance and application of the ICF-CY framework by developing the ICF-CY core set for the CP population based on three age groups (264, 281-284). This was described in detail in section 3.1.1 in the literature review. Simeonsson et al. (294) emphasised that reduction of the ICF content to a limited number of codes would be important for a particular purpose/intervention and would increase the utility of the framework in clinical documentation. Therefore, it is essential to develop brief code sets that are applicable within the context of lower limb orthopaedic surgery in order to document function after surgery, both in research and in clinical practice.

Chapter 5 Research Setting and Design

This chapter explains the project setting and discusses the methodologies that were used throughout the work described in this thesis. These methodologies were based on the recommendations of the COMET and COSMIN initiatives (10, 11). The choice of these methodologies was also informed by the recommendations made by Williams et al. (215) regarding the important issues that should be considered during developing a COS. These recommendations were: 1) the scope of the proposed core sets should be defined; 2) there should be Patient and Public Involvement (PPI); 3) all stakeholders should be involved; 4) a conceptual framework of the proposed COS should be drawn up; and 5) a steering committee should be set up with a defined role. This chapter also discusses the project registration and provides a description of the study design. This chapter was adopted and extracted from the published paper (295).

5.1 Establishing the scope of the COS

According to the Core Outcome Set-STAndards for Development (COS-STAD) (272), the scope of the COS refers to the setting in which the COS is to be applied, and the health conditions, target populations, and interventions it will cover. It is thought that identification of the scope of the COS at the beginning of the research reduces the risk of subsequent ambiguity of purpose arising. In addition, a clear definition of the scope will help potential users to decide on the relevance of the COS to their work.

The scope of the proposed COS is to serve as an international standard that can be used to evaluate the overall success of lower limb orthopaedic surgery among ambulant children and young people with CP. It will comprise important outcomes (domains) and outcome measures that should be used as a minimum in research settings.

5.2 Patient and Public Involvement

There is increasing awareness that the involvement of patients and the public as research partners can benefit the research. In 1996, the NIHR supported the establishment of the INVOLVE initiative to support patient involvement as research partners in National Health Service, public health and social-care research (296). According to INVOLVE, PPI requires patients to be involved in designing the study, and the research is carried out ‘with’ or ‘by’ members of the public (296).

Two individuals contributed to this study and provided PPI input and advice: an adult with CP who has previously undergone lower limb orthopaedic surgery and a parent of a child with CP, who also represents a group of approximately 40 families of children with CP. Consultation with these two PPI partners occurred in a variety of settings, including face-to-face meetings and email correspondence. The two PPI partners received an explanation of the research problem and research design details were discussed. They confirmed the importance of the topic from the patient/parent perspective and partnered in the design of the study. In particular, they helped with the information material to support the interviews with caregivers and children that would form the basis of the research and the Delphi methods. As a result, the study design was refined, tips were given on how to maximise the size of the sample of participants (caregivers and children) and feedback was offered on the study materials, such as the patient information sheets and the interview topic guide questions.

5.3 Stakeholder involvement

The involvement of multiple stakeholders in the consensus process for a COS is also strongly recommended by the COMET guidelines (10) and COS methodologists (9, 297). Key stakeholders are recommended to be identified and invited to participate in deciding which outcomes should be included in the COS. Stakeholders may participate in the identification of possible outcomes and/or in consensus exercises to prioritise the contents of the COS. According to COS-STAD (272), key stakeholders should almost always include patients and healthcare professionals who have experience of the clinical condition, but the group may also include regulators, industry representatives and researchers.

As discussed in the literature review, a growing body of evidence highlights that health outcomes and healthcare priorities may differ between stakeholder groups, for example, between researchers, clinicians, parents and children. These differences in opinion highlight the need to ensure that multiple stakeholder groups contribute to the COS development. Patient involvement in outcome selection was initially proposed at an OMERACT meeting in 2000, when clinicians struggled to answer the question of what would be defined as a ‘clinically important change’ in response to treatment. New outcomes that were of significant value to patients were identified and integrated into the core outcome set for rheumatoid arthritis clinical trials (259).

Similarly, the JLA identified the priorities of children involved in paediatric surgical intervention for lower limb conditions and discovered that these priorities were different from those that were identified

through a Delphi process involving paediatric orthopaedic surgeons (292, 298). These differences highlight the importance of involving all relevant stakeholders when considering research priorities.

However, including only children and young people who undergo the treatment does not provide us with information on which specific aspects families rate as most important, and whether there are differences between families in the aspects that they find important. Since families are unique, it can be expected that families differ in what they consider important in the care of their children. Knowledge of what parents consider to be important should provide us with more insight into individual differences related to satisfaction with services and might be helpful in the implementation of family-centred services.

Therefore, the two main groups of stakeholders invited to participate in this COS development are outlined in Table 14.

Table 14 Stakeholder groups

Stakeholder group	Sub-group	Criteria
Healthcare professionals and researchers	Healthcare researchers, trialists	- Professionals who worked in any field of clinical research that was relevant to lower limb orthopaedic surgery (e.g., lower limb surgery, pre- or post-operative rehabilitation, gait-related data) - Methodologists who worked during the project as researchers in this field - All researchers involved in this project were required to be authors of published scientific articles on clinical research in CP
	Healthcare professionals (clinicians)	- Clinicians from different disciplines (i.e., orthopaedics surgeons, physiotherapists, occupational therapists, gait lab clinicians) - Had clinical experience of the management of children and young people with CP, and in particular, management of children who had lower limb deformities.
Children and parents (caregivers)	Children and young people with CP	- Ambulant children [age 8-18 years old] with CP - Had undergone or were being considered for orthopaedic surgery to address lower limb musculoskeletal deformity and/or gait-related pathology
	Parents (caregivers)	- Carers or family members who had a close relationship and lived experience of a child who fulfilled the above criteria. - The term “Parents” was used throughout this thesis which reflect children’s representatives from family members or caregivers

5.4 A conceptual framework

The COMET initiative emphasised the need for a comprehensive framework of healthcare when developing a COS to provide essential structure to the conceptualisation of domains (10). Several frameworks have been developed to date that classify health, disease and outcomes (Table 15).

Table 15 Health outcomes frameworks

Framework	Focus	Reference
OMERACT	Comprises three core areas: death, life impact and pathophysiological manifestations. These core areas are further categorised into core domains	(255, 299)
Outcome Measures Framework (OMF)	Comprises six outcome domains: survival, disease response, events of interest, patient/caregiver-reported outcomes, clinician-reported outcomes and health-system utilisation	(300)
Patient-Reported Outcomes Measurement Information System (PROMIS)	Comprises three main domains: physical (symptoms and functions), mental (effect, behaviour and cognition) and social wellbeing (relationships and function)	(301)
WHO	Comprises three broad health domains: physical, mental and social wellbeing	(302)

The ICF-CY framework (detailed in section 3.1.1) has been used as a conceptual framework and classification tool in the development of the proposed COS. The ICF-CY has been widely used in CP research as a means to describe communicative functioning, disability and the impact of contextual factors, to classify the content of assessments and outcome instruments and to analyse and compare data (135, 136, 251, 262, 285, 303, 304). Viehweger et al. (142) reported that paediatric orthopaedic evaluations would be improved through the use of different measurement tools for each ICF-CY domain alongside specific QoL measurement tools. Therefore, the suggested COS will include both the ICF-CY framework and other outcomes that do not fit precisely in the ICF-CY framework, such as QoL outcomes.

In the present study, the ICF-CY framework will be used to:

1) Categorise concerns and experiences of children and parents on the surgical outcomes.
2) Link the surgical outcome domains and outcome measures used in the literature.
3) Explore stakeholders' perspectives regarding the key health areas post-surgery.
4) Inform stakeholders about COSs.

Hence, the ICF coding and linking rules will be used as a method of data analysis. In this approach, information is linked to ICF-CY codes and categories though the use of standard linking rules (305). This

provides a common taxonomy through which data can be compared within and across studies. ICF-CY linking rules will be detailed in the next relevant chapters.

5.5 Steering committee

A national steering committee was formed to initiate and support the development of this COS. Members of the committee were chosen to represent various disciplines and types of expertise. As a result, the committee comprised ten members: five health professionals (clinicians and researchers) from different disciplines with ample experience in CP orthopaedic management and clinical research; two experts in consensus-based research procedures; two children's representatives; and one parent representative.

Engagement with the steering group took place remotely via email. The involvement of the steering group will be detailed in relevant chapters. The main role of the steering group was to discuss and advise on all steps of the COS development process, particularly the processing and finalisation of the outcome domains. The terms of reference of the steering committee are presented in Appendix 1.

5.6 Project registration

This project was registered with the COMET database in 2018, with the registration number 1236 (306).

5.7 Study protocol

There are potential sources of bias in the COS development process. In its handbook, COMET recommends that a protocol should be developed prior to the start of the study and made publicly available. In order to improve transparency and share methods with others, the protocol for this study was published in an open-access journal (295).

5.8 Study design

The research design followed COMET and COSMIN recommendations (10, 11). It comprised of six main studies to address the research aims and objectives. Figure 9 illustrates the approach that was taken specifically to develop a COS for lower limb orthopaedic surgery in ambulant children and young people with CP.

Study I Qualitative Evidence Synthesis. A review of the qualitative literature in this area of research was undertaken. This study aimed to identify the health outcomes that are key for children with CP and their parents in relation to lower limb surgery. It also aimed to understand the experiences of children and their families pre- and post-surgery, and used the ICF-CY as a reference. The research question for this study was: from the perspective of children with CP and their parents, what is the experience and expectation of outcomes after lower limb surgery?

Study II Scoping review. This study aimed to summarise and describe the health domains related to lower limb surgery in CP, as assessed in the published literature and describe the outcome measures used in the identified studies, using the ICF-CY as a reference. The research questions for this study were: what outcomes are reported in the medical literature after lower limb orthopaedic surgery for ambulant children with CP? What outcome measures are used in this field?

Study III Qualitative study. This study was set up to identify outcome domains that are important to children and young people with CP, their parents and health professionals, and to quantify these characteristics using the ICF-CY coding system. The research question for this study was: what outcome domains are considered important by ambulant children and young people with CP, their parents and health professionals in relation to lower limb orthopaedic surgery?

Study IV Delphi survey study. This study aimed to develop consensus from international CP experts as well as children and young people with CP and their parents, regarding the most important outcome domains. The research question for this study was: which outcomes do children and young people with CP, parents and healthcare professionals think should be included in a COS for lower limb orthopaedic surgery?

Study V Consensus meeting. This study aimed to obtain a final consensus on the outcomes identified in study IV by selecting the most relevant outcome domains to be included in the core set.

Study VI Selection of optimal outcome measures. This study aimed to critically appraise used outcome measures and obtain a final consensus on the optimal outcome measures for each identified health area in the core set.

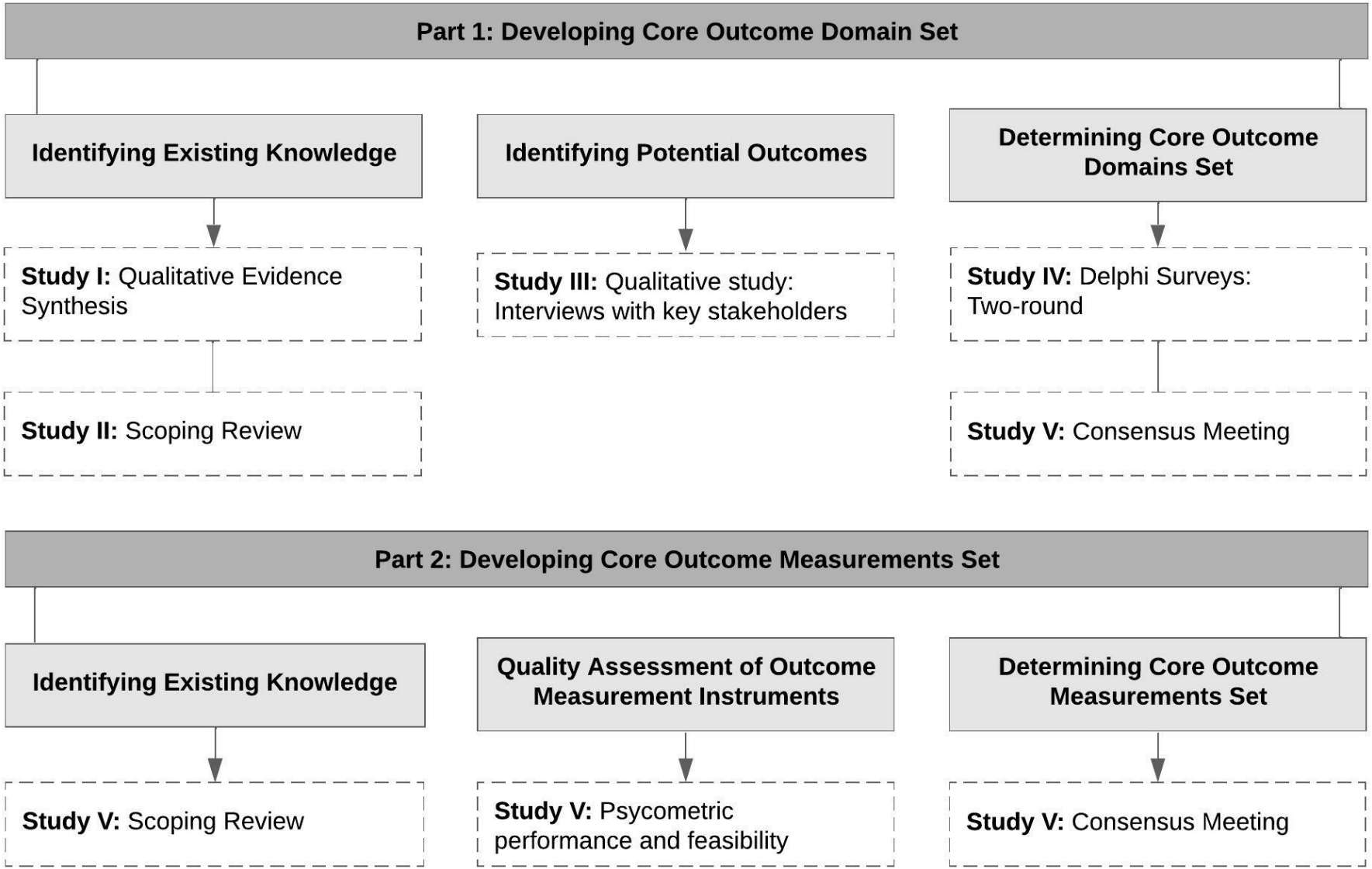


Figure 9 Project study design

Chapter 6 Qualitative Evidence Synthesis

Patients' and parents' views about lower limb orthopaedic surgery for ambulant children and young people with cerebral palsy: a qualitative evidence synthesis

This chapter describes the scoping search, methods and approaches used for the Qualitative Evidence Synthesis (QES). This is the first methodological strand to the research in this thesis. The outcomes identified in this chapter will be carried through to a consensus process to ratify the outcomes to be included in the final COS. This chapter was adopted and extracted from a published paper (307).

6.1 Background

As discussed in the introduction to this thesis, consideration of the patient and family's experiences is increasingly recognised worldwide as one of the elements of high-quality healthcare. The development of a COS that includes the views of young people and parents would address the issues of unmet expectations and inconsistency of experiences (10). Qualitative research can highlight important issues that are relevant to particular populations, and this can support healthcare decision-making. Armstrong et al. (308) suggested that interventions and outcomes that were important to patients could be selected based on qualitative findings. Conducting a QES, which would involve the synthesis of several primary qualitative studies, and that was focused on the experiences of ambulatory children with CP would have three benefits:

First, as part of a large study on the development of a COS for orthopaedic surgical interventions in ambulatory children with CP, it would provide a comprehensive understanding of the experiences of children and young people with CP who have undergone lower limb surgery and could offer a preliminary list of important outcomes for consideration.

Second, information gathered from a QES would increase the volume, depth and diversity of stakeholder perspectives that would be incorporated into the COS (309). This is important because perceptions among stakeholders could vary widely (210, 310). Inclusion in the QES of the evidence from a wider range of participants would provide a better understanding of the issues than were available at the time of the project and make an important contribution to the goal of providing better services.

Third, it could therefore contribute to the COS development process through the provision of a list of surgical outcomes to be considered (10, 254). Several reviews of the clinical trials reflect the researchers' and clinicians' perspectives of the outcomes but not those of patients or parents. Thus, it was important not to limit the synthesis to one group of participants.

Previous QESs that were undertaken as a part of COS development in critical type 2 diabetes, bariatric surgery and critical illness identified outcomes of importance that had not been reported in systematic reviews of clinical trials (311-314). For example, Gorst et al. (313) found several outcomes from a QES of type 2 diabetes treatment that differed from clinical trials outcomes, such life impact outcomes.

Scoping search

A scoping search was carried out to identify existing QESs and establish the need for an additional review. The search and results are illustrated in Appendix 2. However, the search did not identify any QESs that had explored the experiences of lower limb orthopaedic surgical interventions.

Therefore, the aims of this QES were to:

1. identify and synthesise qualitative research that explored the experiences of children and young people with CP who had undergone lower limb orthopaedic surgery, and the experiences of their families,
2. identify health outcomes related to lower limb orthopaedic interventions that are considered important from the perspectives of ambulatory children and young people with CP and their parents.

Protocol and Registration

The review protocol was registered with PROSPERO with the registration ID CRD42018089538.

This study is reported in accordance with the Enhancing Transparency in Reporting the Synthesis of Qualitative Research (ENTREQ) guidelines (315) and the statement supplemented by the list of Preferred Reporting Items for Systematic reviews and Meta-Analyses (PRISMA) (316).

6.2 Methods

Best-Fit Framework methodology

Qualitative research can be synthesised in several ways, including *integrative/summative*, through which data are pooled and assembled, and *interpretive* synthesis, through which concepts and theories are developed (317-319). Different methods adopt different approaches (318, 319). Examples of different approaches are shown in Table 16. Booth (2017) suggests several factors that should guide the selection of the synthesis method: for example, the type of review that is being undertaken and its scope, the review question(s) to be answered, and the aims of the review and existing evidence (320).

In this study, the primary aim was to gather various perspectives and experiences that could contribute to the creation of the COS for orthopaedic surgical interventions for children with CP, rather than to produce an explanatory model or theory. Therefore, a summative approach that enabled the concurrent consideration of studies that employed a range of methods (e.g., deductive or inductive analysis) was appropriate. However, it was anticipated that the synthesis would generate evidence that would extend beyond the chosen conceptual framework (i.e., ICF-CY) and could not be fitted easily into the framework, therefore, interpretation and translation of this evidence into outcomes was also likely to be necessary.

For this reason, a Best-Fit Framework was chosen, which used a priori themes from an existing model as a starting point for deductive analysis. The data that could not be accommodated within the framework were analysed inductively, using thematic synthesis (321). The value of this approach was that it combined both framework and thematic synthesis.

The Best-Fit Framework involves seven steps, which are shown in Table 17 (321). The QES that was used in this case employed steps one to five. Steps six and seven were omitted because they are designed to produce a conceptual interpretation, which was not required for this QES. This synthesis involved the description and integration of available data.

Table 16 Different Qualitative Evidence Methods

Methods	Description	Approach
Meta-ethnography	Reciprocal translational analysis (RTA) identifies key themes in each study, then seeks to translate these into the context of each study. It may then extent these translations into a higher order interpretation; a broader picture of the phenomenon	Interpretive
Thematic analysis	Identification of major / recurrent themes in the studies identified; summary of findings of primary studies under these thematic headings.	Interpretive/ Integrative
Framework/Best-Fit Framework	Evidence from primary studies coded against the themes of a priori framework and new themes are generated from evidence not captured by the a priori framework.	Interpretive/ Integrative
Grounded theory	Constant comparative method identifies patterns and interrelations in primary data. Sampling responds to analysis, until theoretical saturation reached.	Interpretive

Table 17 Summary of 'best-fit' framework synthesis approach

Step	Description	
Step 1	Define review question	
Step 2	Identify relevant best-fit framework	Systemically identify relevant primary research studies
Step 3	Conduct quality assessment	Extract data from primary included study
Step 4	Code evidence from included studies into a priori framework identified in step 2	
Step 5	Create new themes by performing thematic analysis on any evidence that cannot be coded into a priori framework	
Step 6	Produce new framework composed of a priori and new themes supported by the evidence	
Step 7	Revisit evidence to explore relationships between themes in order to create a model	

6.2.1 Step one: The review questions

The research question was formulated by using a series of considerations: Sample, Phenomenon of Interest, Design, Evaluation, and Research type (SPIDER) (322). Table 18 shows ways in which SPIDER elements were used in this QES.

Table 18 SPIDER research question

SPIDER elements	Description
Sample	Ambulatory children and young people with CP and their parents
Phenomenon of interest	The experience of surgery and the key aspects of health
Design of the included studies	In-depth qualitative interviews (focus groups and individual interviews)
Evaluation	The participants' experiences and perspectives
Research type	Qualitative research only

The SPIDER question was formulated as follows: what are the experiences and expectations of children and young people with CP and their parents regarding outcomes after lower limb orthopaedic surgery?

6.2.2 Step two: Search and Study selection

6.2.2.1 Searching

For the purpose of the Best-Fit Framework, it was necessary to identify (A) a relevant framework and (B) the relevant primary studies that were to be included in the review.

A. Identification of a relevant framework

The ICF-CY framework was selected as the best-fit model to synthesise the data from the primary studies for two reasons: the use of the ICF-CY terminology could be linked to previous studies in CP to explore the concept of orthopaedic interventions, and the themes generated represented a robust base of knowledge that formed the first list of a future ICF-CY COS for lower limb orthopaedic surgery of ambulant children with CP.

B. Identification of relevant primary studies

A comprehensive search was conducted through five databases: the biomedical site Embase; the US National Library of Medicine site MEDLINE; the biomedical and life sciences database PubMed; the cumulative index of nursing and allied health literature (CINAHL); and the American Psychological Association's collection PsycINFO, from inception to 11 April 2020. Appendix 3 lists the keywords that were used in the search, their combinations and the references that were obtained from every database. To identify further studies, a supplementary search of the references of every identified study was applied. A Web of Science citation search was conducted to screen the references of all the studies that were of direct relevance to the synthesis.

6.2.2.2 Study selection

The criteria established in the review protocol served as the basis for which the studies that were identified from the databases were included or excluded in the review. This process involved screening the title and abstract first and then screening the entire text. Selection of studies was based on the pre-identified inclusion and exclusion criteria of the studies' characteristics as shown in Table 19.

Table 19 Inclusion and Exclusion criteria

Inclusion Criteria
- The study population comprised individuals who had been diagnosed with CP and/or their parents
- The participants were considered ambulatory or had been classified as within level I, II, III of the GMFCS
- The participants had experienced orthopaedic interventions for lower limb conditions
- The study employed qualitative design, data collection and analysis
- In mixed-method studies, data from the quantitative and qualitative approaches had been analysed and reported separately
- The full study had been published in English
Exclusion Criteria
- Opinions, editorials
- Studies that contained quantitative data only
- Studies that involved populations that represented several types of disability (if the authors had not defined findings by type or specified which quotes were linked to individuals with CP)
- studies that comprised only an abstract or were published as posters, since these abstracts had not been peer-reviewed and could have contained preliminary results rather than final analyses, which would limit the author's ability to assess their relevance adequately

At least two researchers conducted this process to ensure transparency and reproducibility (323). The researcher (HA) screened the titles and abstracts of the identified studies. To achieve certainty regarding which studies should be included and excluded, another member of the research team (FT) screened titles and abstracts for a random sample of the identified studies. Studies that were potentially relevant were checked based on a full-text reading by HA. Studies that HA deemed should be included were checked by FT through full-text screening. Studies that HA deemed should be excluded were sent to two other research team members (TT and HD) for review of the titles and abstracts. If TT or HD was uncertain as to whether a study should be included after they had read the title and abstract, the study was returned to HA or FT for further screening. If they remained uncertain about inclusion, the full text was checked again by the researcher (HA). Any ambiguities or uncertainties were resolved through research team discussion.

6.2.3 Step three: Quality Appraisal and Data extraction

6.2.3.1 Quality Appraisal

Hannes et al. (324) argue that, in order to differentiate between studies of high- and low-quality, all qualitative evidence reviews must include critical appraisal. Various tools are available to appraise the quality of qualitative research (Table 20).

Table 20 Quality appraisal tools

Tools	Reference
Critical Appraisal Skills Programme (CASP) Checklist	**(325)**
Joanna Brigg's Institute's Qualitative Assessment and Review Instrument (JBI-QARI)	**(326)**
Qualitative Research Review Guidelines- Relevance, Appropriateness, Transparency and Soundness Of Approach (RATS)	**(327)**
Standards For Reporting Qualitative Research (SRQR)	**(328)**
Consolidated Criteria for Reporting Qualitative Research (COREQ)	**(329)**
Evaluation Tool for Qualitative Studies (ETQS)	**(330)**

The CASP checklist was chosen as the best fit tool for quality assessment in this case for the following reasons: it is easy to administer; it is a time-saving tool since it contains only ten questions; it is the tool most commonly used in health-related QES in Cochrane and WHO guideline processes (331); and the CASP checklist aligns with the recent Cochrane guideline on the assessment of the methodological strengths and limitations of qualitative studies (331). The ten questions that comprise the CASP are shown in Table 21.

In the context of the evaluation process, each question of the CASP can be answered as 'yes', 'no', or 'can't tell'. Although the CASP was not designed to provide a numerical score, some authors have assigned numerical scores to each question, as illustrated in Table 22. This practice has been used in previous qualitative evidence syntheses, in which each question was scored as 1 if the answer was recorded as 'no', 2 if recorded as 'can't tell' or 3 if recorded as 'yes' (332, 333). This method gave each study a score that ranged from 10 to 30.

Table 21 The CASP checklist

CASP Checklist
Q 1: Was there a clear statement of the aims of the research?
Q2: Is a qualitative methodology appropriate?
Q3: Was the research design appropriate to address the aims of the research?
Q4: Was the recruitment strategy appropriate to the aims of the research?
Q5: Was the data collected in a way that addressed the research issue?
Q6: Has the relationship between researcher and participants been adequately considered?
Q7: Have ethical issues been taken into consideration?
Q8: Was the data analysis sufficiently rigorous?
Q9: Is there a clear statement of findings?
Q10: How valuable is the research?

Table 22 Level of quality assessment

Level	Aggregate Score	Score
Yes	been extensively addressed	3
Can't tell	been addressed partially	2
No	not been addressed	1

Dixon-Woods et al. (334) appraised studies for quality and relevance in five categories as presented in Table 23. This categorisation was also used to determine which studies should be included in the present review (335). Studies that fell into categories one and two were included, categories four or five were excluded and studies in category three (unclear) were discussed with the research team to reach a consensus on whether to include or exclude them.

Table 23 Dixon-Wood criteria for quality and relevance

Category	Definition
1. Key paper	Conceptually rich and could make an important contribution to the synthesis
2. Satisfactory	Satisfactory insight into the phenomenon
3. Unclear	Required further screening
4. Fatally flawed	Flawed in terms of transparency of methodological procedures
5. Irrelevant	Not addressing the review question or aim

There is no consensus about the use of quality appraisal tools in qualitative research. Some reviewers recommend the exclusion of low-quality studies as they do not make meaningful contributions to the analysis. Others argue that quality appraisal tools do not measure quality (336). Sandelowski et al. (337) argued that some qualitative researchers might not have reported all methodological and procedural details in the published paper, and that the absence of such details from a study report do not necessarily indicate their absence from the research process. In addition to these reasons, as the review aim was to aggregate and summarise all available findings, rather than to formulate new theories, studies were not excluded at this stage based on the CASP findings. However, quality assessment was used to moderate the impact of the review findings based upon the quality of the included studies.

It is recommended for this process to be conducted by two researchers so that ambiguities can be addressed, and certainties confirmed (324, 331). At least two researchers appraised the quality of the studies and met to discuss their decisions.

6.2.3.2 Data Extraction

As recommended by Noyes et al., (331), the contextual, methodological and analytical data of each included study were extracted and reported. The element of each data is shown in Table 24. A template was created for the extraction of these data from the identified studies. The data extraction table was pilot-tested by HA on a sample of relevant studies and agreed upon with a qualitative synthesis expert (FT).

Table 24 Data extraction element

Type	Element
Contextual	Study setting, population, participant characteristics, and the intervention that had been delivered.
Methodological	Methodological design and the approach taken in the study; methods that were used to decide who should be recruited to take part and how; and the methods that were employed for data collection and analysis.
Analytical	The quality assessment criteria that were applied to each study.

Approach to synthesis

Two methods were employed to identify major themes and to examine the studies in a structured, yet flexible manner. First, an a priori framework that incorporated ICF-CY components was used as an existing model to facilitate data synthesis (Step four). Second, data that did not fit the ICF-CY framework were analysed thematically (338) (Step five).

An Excel sheet template was used to aid data managements and synthesis. Two researchers, trained in ICF-CY coding (HA and JP), independently checked the assignment of key findings from the included papers into the appropriate section of the ICF-CY framework. All stages of the thematic synthesis were also examined by HA, JP, and FT to ensure the consistency in the interpretations. Any disagreements were discussed by the same researchers.

6.2.4 Step four: Coding of evidence against the ICF framework

In order to link data to precise ICF codes, a set of standardised linking rules has been created, utilised, and researched. The first set of linking rules was created in 2002 (339); it was adjusted and modified in 2005 (340), 2016 and 2019, when the linking rules were refined and standardised (305, 341). In this study, the researcher chose to use the most recently updated (2019) linking rules (305), which served as

the starting point of the synthesis. Figure 10 shows the process by which each concept was linked to the framework. The 2019 update advised utilisation of ten linking rules, as shown in Table 25.

For example, if the primary study contained a quote “I had trouble walking”, the concept that would be extracted from this statement was walking. Walking was linked to the first ICF-CY level ‘activity and participation’, the second ICF-CY level ‘d4-mobility’ and to the more precise third level category ‘d450-walking’. Any data from the included studies accorded with eighth and tenth rules were moved to a second thematic synthesis. For example, meaningful concepts that referred to ‘quality of life; QOL’ were assigned to the ‘not definable- quality of life’ category, which was then analysed thematically.

Table 25 ICF linking rules

Number	Rule
1	Acquire good knowledge of the conceptual and taxonomical fundamentals of the ICF, as well as of the chapters, domains, and categories of the detailed classification, including definitions before starting to link meaningful concepts to the ICF categories
2	Identify the purpose of the information to be linked by answering the question ‘what is this piece of information about?’ or ‘what is this item about?’. The answer to these questions will help to identify the main concept(s) most relevant to be linked to the ICF
3	Identify any additional concepts contained in the piece of information in addition to the main concept(s) already identified in the previous step
4	Identify and document the perspective taken within a certain piece of information when linking it to the ICF
5	Identify and document the categorisation of the response options
6	Link all meaningful concepts, the most relevant and additional ones, to the most precise ICF category
7	Use ‘‘other specified’’ or ‘‘unspecified’’ ICF categories as appropriate
8	If the information provided by the meaningful concept is not sufficient for making a decision about the most precise ICF category, assign the concept to not definable (ND)
9	If the meaningful concept is not contained in the ICF, but is clearly a personal factor as defined in the ICF, assign the meaningful concept to personal factors (PF)
10	If the meaningful concept is not contained in the ICF, assign this meaningful concept to not covered (NC)

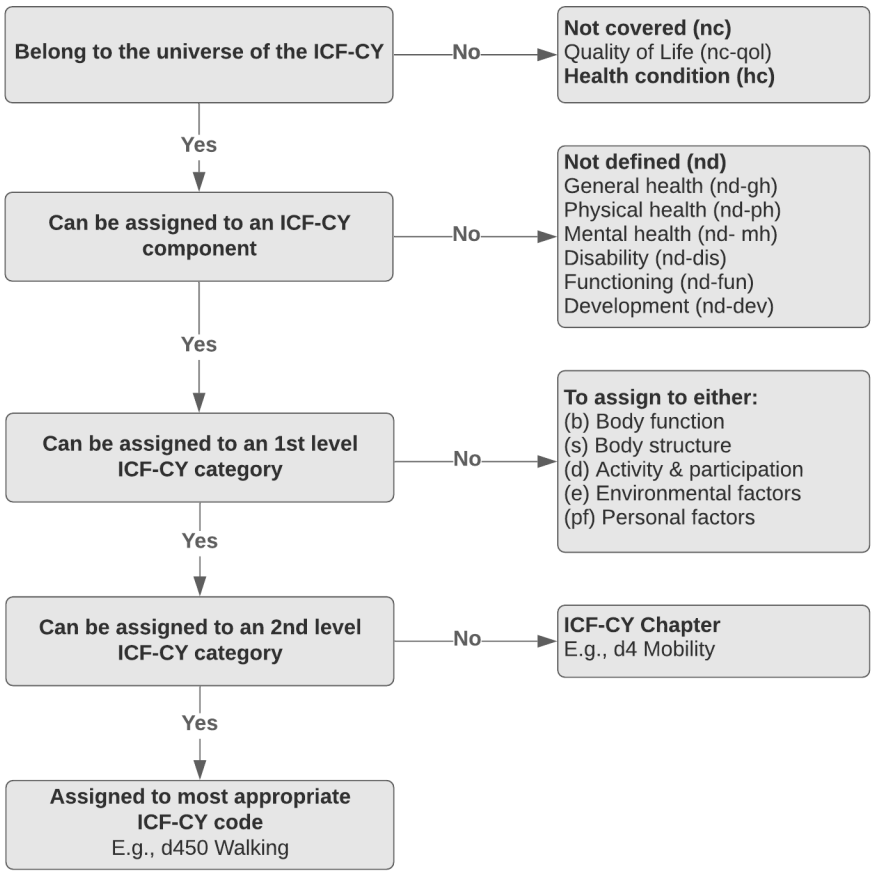


Figure 10 Linking decision tree

6.2.5 Step five: Thematic synthesis

A thematic synthesis was conducted in cases where the data did not fit into the framework. Table 26 indicates the three stages of the thematic synthesis of primary studies, as suggested by Thomas and colleagues in 2008 (338). This method employed an inductive, constant comparison approach to the generation of codes, categories, and themes. This approach is particularly relevant for reviews that seek to shape the outcomes of interventions.

Table 26 Stages used in the thematic synthesis

Stages	Process
Stage 1	Coding of text line-by-line
Stage 2	Condensing of similar study findings into descriptive themes: a stage of interpretation that remains 'close' to the primary studies
Stage 3	Generation of analytical themes: a stage of interpretation in which the reviewers 'go beyond' the primary studies and generate new interpretive constructs, explanations, or hypotheses – that is, theory about the importance of treatment outcomes

Line-by-line coding

Data from studies that were assigned under the eighth and tenth rules were read and then each sentence of text was coded to distill its meaning. At least one code was applied to every sentence. Each new code that was assigned to a statement was added to the 'codes bank', and new codes were developed when necessary. All identified codes were defined to facilitate the research team's understanding of the meaning of the code.

Development categories

At this stage, codes were organised into categories to capture the meaning of groups of initial codes. All identified categories were translated into concepts to facilitate the research team's understanding of the meaning of the category.

Generation of overarching themes

Themes were generated to capture the data that involved similar concepts from the developed categories. These categories referred to both the desired outcomes and the actual experiences that children or parents described in relation to those interventions concerning musculoskeletal deformities of the lower limb. At this stage, three researchers (HA, JP, and FT) independently linked overarching themes of outcome domains to lower limb surgery.

6.3 Results

6.3.1 Searching strategy

The literature search identified 8,252 studies. A manual search of the references that were cited in the available reviews of CP yielded an additional nine studies. All identified studies were imported into an EndNote library (Endnote X8), duplicates (6,123) were removed, and the remaining 2,138 title/abstracts were screened. A total of 26 were selected for full-text review. Of these, 20 were excluded: eight were grey literature (conference abstracts) (342-349), five did not provide data regarding ambulatory CP (284, 350-353), five did not provide qualitative data (354-358), one was based on the perspectives of the adult CP population (359), and the data for one study were unavailable (360). Therefore, the synthesis was of six studies that considered the experiences and perspectives of children and young people with CP and their parents regarding lower limb orthopaedic surgery. The search process details are outlined in Figure 11.

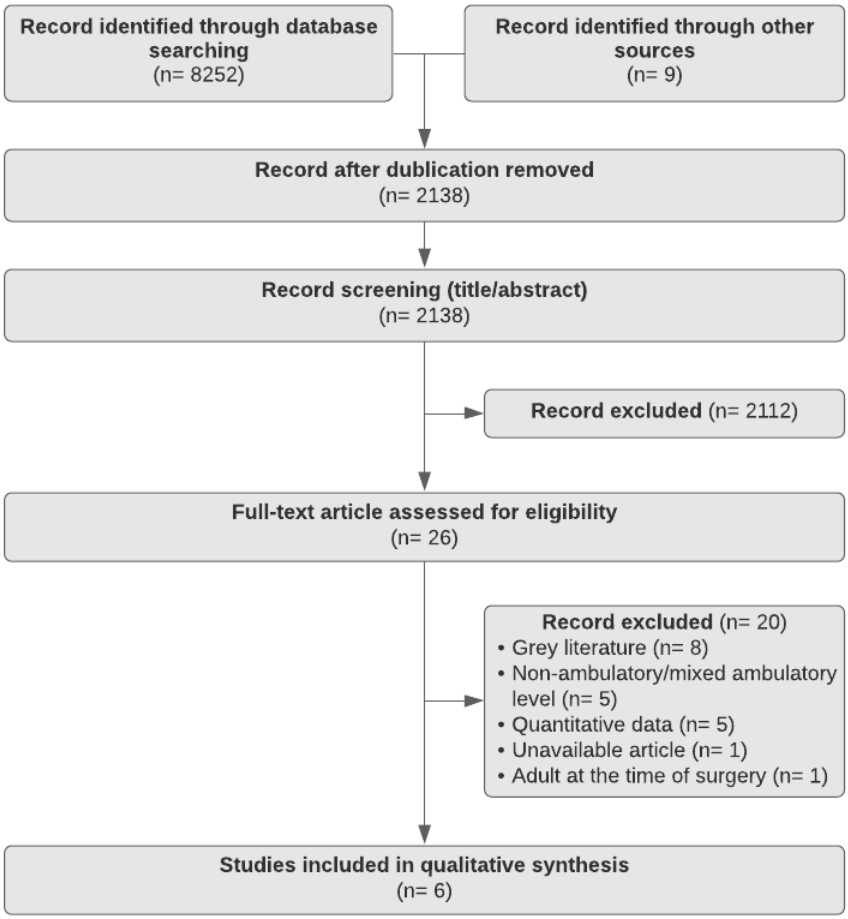


Figure 11 Flowchart of the study selection process

6.3.2 Included studies

Table 27 provides a summary of the characteristics of the included studies. The studies synthesised findings from primary qualitative research that had been conducted in three countries; four studies were conducted in Norway, one in Finland, and one in France. Sample sizes ranged from seven to twelve participants. One study interviewed children and young people; one study interviewed parents only. Four studies reported results from both parents and individuals with CP. Of these four studies, two reported the same population at different stages of surgery (i.e., pre- and post-surgery). The age of the participants in the studies ranged from eight to twenty-two years old. Most studies included children's and parents' views following lower limb surgery (n= 5); Only one study reported pre-surgery perspectives. None of the studies were conducted to specifically examine children's or parents' priorities. All the study designs involved semi-structured interviews.

6.3.3 Quality appraisal

The CASP score ranged between 25 and 29 regarding aspects such as design, recruitment, data collection, rigour, data analysis and relevance of the findings. All the studies offered limited descriptions of the relationships between the researchers and the participants and of ethical issues.

Table 28 shows the scores and ranks that HA and JP gave to all papers during appraisal. Disagreement was due to the unclear reporting of ethical considerations and the limited rigour of the analysis descriptions. HA and FT agreed that all six studies were at least satisfactory, and HA felt that three of these were key papers (Table 28). Table 29 shows the final findings of the critical appraisal of the studies as scored through use of the CASP tool.

Table 27 Characteristics of included studies

Study	Country	Participants	Children age	Methods	Data analysis	Study aim
(Capjon and Bjørk, 2010)	Norway	8 parents and children	12 -16 years	Semi-structured interview	Kvale's method	Explores post-operative family situation, rehabilitation, and interdisciplinary cooperation for ambulant children with cerebral palsy after multilevel surgery
(Capjon and Bjork, 2010)	Norway	8 parents and children	12 - 17 years	Semi-structured interview	Kvale's method	Explores the pre-operative situation of children accepted for multilevel surgery for cerebral palsy and their parents
(Iversen, Graue, and Clare, 2009)	Norway	12 parents	8 - 16 years	Semi-structured interview	Phenomenology	Explores parents' experiences of their child with cerebral palsy going through a surgical procedure
(Lehtonen, Maenpaa, and Piirainen, 2015)	Finland	10 young adult	15 - 22 years	Semi-structured interview	Phenomenology	Examines the perception of adolescents concerning the results of surgery on personal physical functioning in the environment five or more years after single-event multilevel surgery
(Hoiness, Capjon, and Lofterod, 2014)	Norway	7 children and parent	9 -16 years	Semi-structured interview	Kvale's method	Investigates children who underwent SEMLS regarding postoperative rehabilitation and pain, gait parameter 1 year after surgery and mobility 5 year after surgery
(Stephan-Carlier, Facione, Speyer, Rumilly, and Paysant, 2014)	France	11 children and parent	13-21 years	Semi-structured interview	Descriptive	Explores the perceptions of parents and the experience of their operated children in terms of quality of life and surgical outcome

Table 28 Team appraisal results

Study	HA CASP Score	HA RANK	JP CASP SCORE	FT RANK	Difference in CASP score	Difference in Dixon-Wood Rank
Capjon et al (2010)	26	KP	28	SAT	2	Different
Capjon et al (2010)	27	KP	27	SAT	0	Different
Iversen et al (2009)	29	SAT	29	SAT	0	Agreed
Lehtonen et al (2015)	28	KP	26	SAT	2	Different
Høiness et al (2014)	25	SAT	24	SAT	1	Different
Stephan-Carlier et al (2014)	26	SAT	26	SAT	0	Agreed

KP: Key Paper **SAT:** Satisfactory

Table 29 Findings of the critical appraisal of the included studies using CASP tool

Study	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	Q10
Capjon et al (2010)	●	●	●	●	●	○	○	●	●	●
Capjon et al (2010)	●	●	●	●	●	○	○	○	●	●
Iversen et al (2009)	●	●	●	●	●	●	○	●	●	●
Lehtonen et al (2015)	●	●	●	●	●	○	●	○	●	●
Høiness et al (2014)	●	●	●	●	○	○	○	○	○	●
Stephan-Carlier et al (2014)	●	●	●	●	●	○	○	○	○	●

● Yes ○ Can't tell

6.3.4 Synthesis findings

The analysis created 26 initial codes related to the ICF-CY framework. These codes were categorised into four ICF-CY themes. The themes were: (1) body function and structure, (2) activity and participation, (3) environmental factors and (4) personal factors. Four codes that did not fit the ICF-CY framework created two additional themes: (5) emotional well-being and (6) goal setting.

The distribution of the studies according to each ICF-CY concept and each of the two additional themes is shown in Appendix 4. A list of the identified outcomes is summarised in Table 30. The outcomes presented in Table 30 were compiled and consolidated from the text.

Table 30 Desired outcomes

Children and young people with CP	Family member	
	For their children	**For themselves**
Theme one: Body function and structure		
Alleviating pain	Alleviating pain	
To be strong, less contracture	To be strong, less contracture	
Good balance, less fall incident	Good balance	
Better lower-limb alignment	Better lower-limb alignment	
To have better gait appearance	To have better gait appearance	
Theme two: Activity and Participation		
To learn and adapt new skills	To learn and adapt new skills	
Being functionally independent	Being functionally independent	
To walking normally, faster, long distance	To walking normally, faster, long distance	
Be able to move around independently	Be able to move around independently	
Participation with peers	Participation in everyday life	
Feel less isolated		
To be socially accepted		
Theme three: Environmental factors		
To have family, friend support	Better social attitude	To have better communication with health professionals
	To have understanding and improved peer attitudes	Facilitate support with health professionals
	To have better health services	Being informed
Theme four: Personal factors		
Less post-surgery complication	Less post-surgery complication	
Theme five: Parents' emotions and well-being		
		To reduce frustration and stress
		To feel understood by family

Theme one: Body function and structure

All six studies reported treatment experiences related to the 'body function and structure' components of the ICF-CY framework. The data linked to the following three ICF-CY chapters: functions of the cardiovascular and respiratory systems (fatigue); sensory function and pain; and neuromusculoskeletal and movement-related function and structure.

1.1 Functions of the cardiovascular and respiratory systems (fatigue)

Improved endurance was described as an important outcome post-surgery to avoid the consequences of fatigue and impaired levels of energy that had an impact on the child keeping up with their friends.

> *"He has to stay in the classroom during breaks, it costs too much [energy] to go out" (parent) (209)*

However, during rehabilitation immediately post-surgery, fatigue could be exacerbated due to the levels of exertion and demand that were needed.

1.2 Sensory function and pain

Reduced pain was also described as an important outcome post-surgery. Some children reported less pain compared with what they had experienced before surgery; however, others, along with their parents, said they had endured unanticipated pain post-surgery that had negatively impacted their daily lives. Examples of these impacts were limited inclusion in community and school activities, sleepless nights, and depression.

> *"We have gone through a half year of sleeplessness and a nightmare of pain. We could never have imagined that it would be so difficult. We have used sleeping pills and at times our daughter has wanted to die. This has been a tremendous challenge for the entire family" (parent) (211)*
> *"I tried to do the same as the other kids, got knee pains, and ended in a wheelchair for a week" (child) (209)*

Participants described a range of sources of pain after surgery. They included the application of casts post-surgery and limited support for, or adjustment of, facilities at school or home, which increased the pain of movement and that exacerbated fatigue.

> *"He has been through operations before, but never with as much pain and exhaustion as this time. He has lost all energy and initiative. We think a lot more could have been done to alleviate his pain from the start, when he got his cast. The past year has been horrible" (parent) (211)*

Parents reported feeling vulnerable when they saw their children in pain, particularly when their child was hospitalised post-surgery.

1.3 Neuromusculoskeletal and movement-related functions and structure

Neuromusculoskeletal and movement-related function and structure were defined as important post-surgery outcomes from the perspective of children and their parents. They sought improvement in joint range of motion, muscle power and muscle endurance. Parents and children felt that these improvements would lead to restored balance, reduced numbers of falls and improved gait appearance.

> *"My knees were crooked, now it doesn't hurt that much, and I don't fall as often and everything and it looks better" (child) (210)*
>
> *"We're worried about her back, knees and hips. We hope she'll have fewer [neuromusculoskeletal] symptoms in the long run" (parent) (209)*

Some described the return of symptoms, such as muscle tightness and joint contractures, one-year post-surgery.

Theme two: Activity and Participation

Five studies reported experiences after treatment of the young people and their parents that were related to the 'activity and participation' components of the ICF-CY framework. Of the nine chapters under 'activity and participation', the data linked to two: mobility and community, social and civic life.

2.1 Mobility

Improvements in mobility were described as an important outcome post-surgery. Mobility was seen in terms of the ability to maintain a steady posture, for example in standing and walking. Improvement in walking referred to the ability to maintain walking steadily, efficiently, proficiently, effortlessly, and to be able to walk quicker and for longer distances than they had been able to manage before surgery. Acquisition and development of new mobility skills, such as running and moving around, was another important surgical outcome for children and their parents.

> *"Maybe I'll be able to walk a bit more like the others. Maybe" (child) (211)*
>
> *"I scored three goals already; I can stand longer and run more, and I couldn't do that before. I can walk longer distances and feel I am faster; this is the best operation I've ever had" (child) (211)*
>
> *"We have achieved the goals we set for him, better balance and ambulatory functionality. Now he walks on the entire sole of his foot, manages to keep up with the others and has participated in gym during the past six months. His gait is faster and steadier. We are very pleased" (parent) (211)*

Some studies were less positive, for example, there were reports of children losing their ability to walk or eventually requiring a wheelchair.

> *"I have really done my best, but this has involved very much training and a lot of repetition … and I thought I would eventually be able to walk farther, but in fact I walk only shorter distances. I can't ride a bicycle because my knees hurt too much" (child) (211)*

2.2 Community, social and civic life

Children, young people, and their parents all described improved interactions and social participation as an important outcome in these studies. Enhanced participation was reported at three levels: in the school, community and with family activity.

> *"Now I hang around more with the other boys in the class; I couldn't do that before" (child) (211)*
> *"We have achieved the goals we set for him … (he) manages to keep up with the others and has participated in gym during the past six months. …. We are very pleased" (parent) (211)*

In addition, social acceptance was another important goal of undertaking surgery for both children and parents.

> *"Getting friends is my motivation, then I can be together with the other children. They can't be bothered to walk home with me, they'd rather be with the others" (child) (209)*

Theme three: Environmental factors

Four studies reported 'environmental factors' that had enhanced or inhibited positive outcomes. Of the five chapters that occur under 'environmental factors' in the ICF-CY framework, the data were found to link to three: support and relationships, attitudes and services, systems and policies.

3.1 Support and relationships

Health professionals' support was considered to play an important role in the success of treatment and the achievement of goals. Parents viewed this support positively and described it as helpful in terms of building a positive relationship with the health professional pre/post-surgery.

"We feel that we have known the staff at the hospital for many years, and we think that they are the people who know most about these operations. She underwent surgery there" (parent) (211)

Children referred to support in terms of positive interactions during the clinical sessions, and as encouragement from families, friends, peers, and health professionals.

"A lot of responsibility was loaded on me. I need understanding and support" (child) (210)
"I don't want my personal assistant in my work practice. The lady in the cafeteria helps me out" (young person) (210)

In contrast, several parents reported concerns regarding a lack of, or insufficient, information. They criticised clinicians for not listening and for talking in medical terminology that they could not understand fully. They reported impacts of this poor communication on them in terms of feeling uncertain and ambivalent about their desired outcomes post-surgery, and feeling incompetent and powerless. These emotions limited their ability to make appropriate decisions regarding the treatment. Consequently, they sought information and support from other families who had children with similar disabilities.

"Health carers did not give the kind of information we needed, and as parents, we want to get exact information" (parent) (361)
"They talked above our heads and used terms we didn't understand. Ten minutes isn't enough to find out what an operation involves. It wasn't until we got home that the questions started coming" (parent) (209)
"We have lost confidence in the system because the physicians do not listen to us. This involved our own children; it's the children they are talking about, and it is the children they talk condescendingly to, and they also speak condescendingly to us" (parent) (211)

3.2 Attitude

Parents described negative attitudes from school peers, which took the form of physical and verbal bullying, as experiences that had negative effects on their children's well-being. They were worried that surgery could be seen by the child as a way to remove themselves from school.

"She cries almost every day when she comes home from school. She's looking forward to the operation because then she won't have to go to school, and that can't be right" (parent) (209)

3.3 Services, systems and policies

The quality and availability of the required sectors of the healthcare system were factors that either facilitated or inhibited the achievement of good outcomes from surgery. Several parents indicated that several unsatisfactory situations acted as barriers that prevented their children achieving the desired treatment outcomes. These included long follow-up waiting times, the setting of interventions (i.e., at home, in school or in the community), the lack of highly qualified health professionals and the presence of inadequate facilities, such as a lack of wheelchair access to sites.

> *"We need commendation for having persevered over so long a period of time; we don't know if what we are doing is right and we feel that we need guidance. We find it hard to look forward to the next six months without the prospect of any support other than that of our capable but inexperienced physiotherapist" (parent) (211)*

Theme four: Personal factors

Five studies reported individual motivation as a 'personal factor' that might affect the achievement of a good treatment outcome.

Parents and children described being motivated by their goals, such as improved daily life activities and being able to socialise with other children. In contrast, post-surgery complications, unmet needs and negative experiences of previous surgery could be demotivating.

> *"I also can't walk with the shoes and orthopaedic supports because they rub and chafe. I've had physiotherapy and training all my life. This time I feel like I haven't made any progress at all, and I think the whole operation was a stupid mistake" (child) (211)*
>
> *"He has had special orthoses, and they were very good, but gradually they caused blisters and abrasions on the inner side of his foot and the skin hardened ... He prefers to run around without it; it's easiest for him, but we know it's not a good thing" (parent) (211)*

Theme five: Parents' emotions and well-being

One study reported the impact of surgery on parents' 'emotions and well-being'. A strong sense of responsibility and dedication to their children had a profound impact on families who sometimes felt vulnerable and frustrated.

"Sometimes unpredictable tasks every day, and a hospitalisation gives even more tasks, worry and tiredness" (parent) (361)

"I experienced a lot of stress, but you have to be strong" (parent) (361)

Theme six: Goal setting

Two studies reported the goal-setting process as a factor that could enhance the chances of a positive surgical outcome. Although some felt that goal setting should be user-driven, others were uncertain of their own abilities to set goals and saw the healthcare professional as the expert.

"The physiotherapist knows best how I should train" (child) (211)

"She (the physical therapist) lets me decide a lot, it feels really good" (child) (210)

6.4 Discussion

This QES aimed to provide an overview of outcomes of lower limb orthopaedic surgery that were deemed important among ambulant children and young people with CP and their parents, and their experiences that might have affected these outcomes. Through the best-fit synthesis of six studies, eighteen outcomes were of importance to the stakeholders (Table 30). A number of experiences, which included the impact of the surgery, the stress of rehabilitation and the importance of communication and support from health professionals, were highlighted by both parents and young people. The studies also highlighted the emotional impact of the surgery and its profound impact on parents. The findings of the QES suggested that there was a need to introduce a range of measures to capture these important outcomes. There was also need for clear information and support during both preparation and the rehabilitation process following surgery.

The findings of the studies showed that pain, fatigue, and gait abnormalities impacted the ability of the children undergoing surgery to function socially. This may be because the symptoms of CP commonly manifest as limitations of physical function, pain and fatigue that are caused by joint contracture, bone deformities and muscle weakness. These in turn can lead to deterioration in gait and function over time in ambulant children (4). The QES highlighted the interaction between the components of the ICF-CY framework in the desired outcomes of ambulant children and young people with CP. For instance, pain and fatigue, which formed part of the 'body function and structure' domain, affected the children's abilities to socialise with other children. This finding resonates with those of another review of the

patient perspective on rehabilitation services, which found that pain and fatigue restricted the everyday activities of children and young people with CP (362).

There are contrasting assumptions regarding whether activity and participation represent important post-surgical outcomes in CP. Thomason et al. (182) stated that "orthopaedic surgeons have one simple but important tool to bring to the table; that is correction of fixed musculoskeletal deformities. This is the domain in which our contributions and outcomes should be assessed". Narayanan (71) suggested that the focus of lower limb orthopaedic surgery for ambulatory children with disability should be an improvement of the quality of their gait (e.g., to introduce more symmetry or to reduce reliance on walking aids) and optimisation of gait efficiency through pain prevention, and preservation or improvement of physical function, activity and participation. Therefore, key stakeholders, including individuals with CP and their parents, should be involved alongside health professionals during the selection of appropriate outcomes for lower limb surgery in order to enhance understanding of the patients' preferences and values.

Environmental and personal factors were shown to have shaped the stakeholders' experiences of lower limb orthopaedic surgery. For instance, a lack of availability of appropriate equipment or local services to support post-surgery rehabilitation was highlighted as being a barrier to achieving the hoped-for positive effects of surgery. Participants identified the importance of support from and communication with health professionals. Effective communication between patients, families and health professionals is paramount to optimise the partnerships which, in turn, underpin positive outcomes of surgery. This review and other studies that have been conducted within the CP population in different healthcare settings suggest that a hierarchical approach might still be prevalent (362-364). Hence, input from the young people and families might enhance interactions with health professionals and facilitate joint decision-making.

The concept of 'personal factors' is not well defined within the ICF-CY framework. The WHO emphasises the importance of attention to personal factors to achieve the desired outcomes of an intervention (12). Several demotivating factors were identified. These included previous symptoms, past negative experiences of surgery, prolonged rehabilitation before the benefits of surgery had become evident and the requirement for significant effort during the rehabilitation process. Lack of motivation was also recognised as likely to have an impact on treatment adherence and outcomes. This lack of motivation

could be partly attributable to damage that had been caused by CP to the motivation control areas in the brain (365, 366).

A family-centred care approach emphasises the importance of collaboration between families and health professionals in goal-setting and decision-making (367). This approach has been shown to improve outcomes and efficiency of interventions, compared with a lack of collaboration (368). The extent to which the process should involve the child with CP and their family is not clear. In the current QES, children and young people and their families held various opinions of shared goal setting. This variation was also reported in a mixed-method review that investigated the experiences that parents of children with CP had with rehabilitation services (363).

Finally, parents reported overwhelming levels of stress and vulnerability, which were associated particularly with post-surgery hospitalisation, the rehabilitation process and the insufficiency of information and resources. This description of the impact of children's surgery upon parents resonates with the findings of a previous study on the well-being of parents of children with CP. This found that parents' self-perceptions, family functioning and psychological well-being had been affected by the children's disabilities (369). This highlights the need for further psychological support for parents.

Strengths and limitations

This QES had several important strengths. To our knowledge, this was the first QES that had been performed with the use of best–fit synthesis in the CP orthopaedic surgery literature for outcome prioritisation that aimed at COS development. The review was conducted by at least two reviewers at all stages, including the study selection, the data extraction, and the synthesis process.

This review has some limitations. Six studies were included comprising 48 participants, and the majority had been conducted in one country, whereas healthcare services may differ amongst countries. There was a lack of directly relevant evidence; that is, no study had been conducted for the explicit purpose of determining young people's or parents' priorities for orthopaedic treatment outcomes and some of the included studies reported limited qualitative data that could be extracted, which may have influenced the researcher's interpretation.

6.5 Conclusion

This review identified key outcomes (n=18) that should be assessed in clinical and research settings, which are important to children and young people with CP and their parents. The identified outcomes should be considered in the development of a COS for lower limb orthopaedic surgery for ambulant CP.

Next step

The outcomes identified from this synthesis (n=18) represented the perspective of children, young adults and their parents. The scoping search revealed no qualitative studies involving healthcare professionals or researchers. It is possible that relevant surgical outcomes from the perspective of healthcare professionals could be missed, if only this synthesis was conducted. Therefore, a scoping review of quantitative studies in the literature was also conducted and this is presented in the following chapter.

Chapter 7 Scoping Review

Outcome domains and measures following lower limb orthopaedic surgery for ambulant children and young people with cerebral palsy: an updated scoping review

This chapter presents the methods employed and results of a scoping review of primary studies on evaluating lower limb orthopaedic surgical interventions for children with CP, across different time points (short and long-term post-surgery). This is the second methodological strand to the research in this thesis. The outcomes identified in this chapter will be carried through to a consensus process to endorse the outcomes to be included in the final COS. This chapter was adopted and extracted from the published paper (227).

7.1 Background

It is important that outcomes of lower limb orthopaedic surgery, to which many children with CP are subjected, can be assessed accurately. Outcome assessments must consider a child's level of function across multiple domains, such as those across the ICF-CY framework (3). Scoping reviews are an efficient way to identify and summarise existing outcome domains and measures as it enables the health outcomes of interest and outcome measures in the field to be mapped out rapidly (370-374).

Wilson et al. (6) explored outcome measures that were used after lower limb orthopaedic surgery in children and adolescents with CP aged from 0 to 20 years. The researchers investigated 229 studies that had been conducted between 1990 and 2011. In these 229 studies, 32 outcome measures were identified. 91% of the reviewed studies focused on impairment level, whereas only 9% considered activity and participation levels (6). The same researchers carried out a further review of the literature that had been published between January 2012 and December 2015 (191). They found that 81 studies were relevant in this area and 19 measures were identified. The review of the literature published since the original review (6) showed no change in the nature of the outcome measures, the majority of which were focused on the 'body function and structure' domain of the ICF-CY framework.

An updated scoping review in this field of research was considered as part of this project as it would have five benefits.

First, there has been rapid progress in this area. Wilson et al. (6) noted the fast growth in research on the topic of orthopaedic surgery within CP studies; they reported a 46% growth in the number of studies that had been published during the period 2001-2011 compared with the number available before 2000. This difference implied that expansion of the evidence base to include studies that had been published after 2015 was crucial to ensure this the project was up-to-date, and that the review would provide insight and contribute new knowledge to the project results.

Second, the concept of patient-reported outcomes was evolving and a moving toward more patient-centred healthcare. Thus, in synthesising the literature, the researcher could develop a better understanding of the relationship between this shift in concept and health outcomes. Outcome measures had been used among recent evidence.

Third, an examination of Web of Science and Google Scholar on 1 August 2018 showed that the original review had been cited 14 times and read 115 times, based on ResearchGate statistics. These findings implied that it was widely cited and used, which suggested that revising and updating this would be useful.

Fourth, this updated scoping review was part of a larger project, the ultimate goal of which was to develop a COS that could be implemented and formally assessed. The previous review (Chapter 6 Qualitative Evidence Synthesis), which focused on the experiences and outcomes that were important to the children and family stakeholder group is supplemented by aggregating the opinions of previous researchers regarding the outcomes they deemed important to measure.

Finally, considering the growing number of RCTs that are being undertaken in paediatrics (375), an updated scoping review would be a major step towards the development of a systematic review to create a core outcome measure set and to avoid the inappropriate use of unproven measurement instruments (See Chapter 11).

Thus, the scoping review aimed to examine and map the range of key health areas (outcomes) and outcome measures that were used after lower limb orthopaedic surgery of ambulant children and young people with CP. The specific objectives were to:

1. Identify the range and frequency of health domains that have been assessed following lower limb surgery,

2. Identify the range and frequency of use of the available outcome measures,

3. Examine whether the outcome measures discussed in the published literature have changed significantly between the two periods (i.e., before 2016 (6, 191) and after 1 January 2016).

Review Question

The Cochrane Handbook states: "The review question should specify the types of population (participants), types of interventions (and comparisons), and the types of outcomes that are of interest" (376).

The PICO format was used to structure the review question (377). In the PICO acronym, 'P' represents population, patient or problem of clinical interest; 'I' represents intervention; 'C', comparison of treatments or interventions; and 'O', outcome of clinical interest and outcome measures. Table 31 shows ways in which PICO elements were used in this review.

Table 31 PICO research question

PICO elements	Description
Population	Ambulatory children and young people with CP and their parents
Intervention	Lower limb orthopaedic surgery
Comparison	ICF-CY outcomes and outcome measures
Outcome	The frequency of use of outcome measures and the range of key aspects of health (outcomes)

The PICO questions were, therefore: What is the frequency of use of the outcome measures that are employed to evaluate lower limb orthopaedic surgery in ambulant children with CP? Which do they cover, based on ICF-CY framework? How do the results of the outcome measures change from 2016 to 2019?

Protocol

The Cochrane Handbook recommends that a review update should be re-conducted through the use of the same methods that the original review employed (378, 379). Davis et al. (380) suggested that the optimal scoping review is "one that demonstrates procedural and methodological rigor in its application". The development of reporting guidelines for scoping reviews would have helped to ensure

the quality and transparency of the review. Such guidelines did not exist for scoping reviews when Wilson's review (6) was published. Therefore, the researcher assessed the review conducted by Wilson and colleagues with the use of PRISMA extension for Scoping Reviews (PRISMA-ScR) checklist proposed by Tricco et al. (381) as a guide. The researcher concluded that the review had been well conducted in terms of using a valid method and following PRISMA reporting guidelines. Therefore, this review protocol was developed in line with the methodology that had been used in the original scoping review (6).

The protocol for this review was developed through the use of the scoping review methodological framework, the PRISMA-ScR (381) and further refined by the Joanna Briggs Institute (382).

7.2 Methods

Table 32 indicates the steps of the scoping review of primary studies, as suggested by Tricco et al. (381).

Table 32 Scoping review methodology

Step	Description
Step 1	Source and search
Step 2	Eligibility criteria
Step 3	Study selection process
Step 4	Data extraction
Step 5	Analysis

7.2.1 Step one: Source and search

The search for relevant literature was performed for the period between January 2016 and July 2019. In order to optimise the chances of identifying all relevant studies, five electronic medical literature databases were searched: MEDLINE (Ovid and in progress), PubMed, EMBASE, CINAHL and the Cochrane central register of controlled trials.

The key search terms included 'cerebral palsy' AND 'surgical procedures' OR 'surgery' OR 'operative'. The reference lists of all included studies were searched to identify any potentially relevant studies. Appendix 5 shows specific key terms and how they were combined for each database.

7.2.2 Step two: Eligibility criteria

Inclusion and exclusion criteria were pre-defined as shown in Table 33, to enable the screening and study search process.

Table 33 Inclusion and exclusion criteria

Inclusion Criteria
- Any study designs, provided that they reported at least one outcome measure
- Included both children and young people with CP
- The children and young people who were included had been diagnosed with ambulatory CP within levels I to III of the GMFCS
- Participants were aged between 0 and 20 years old
- Participants had undergone any lower limb orthopaedic surgery
- Publication data were confined to the period from January 2016 to July 2019, as the original reviews had covered publications up to December 2015.
- The full study had been published in English
Exclusion Criteria
- Studies were purely observational investigations or qualitative studies, that did not include any outcome measure
- Studies were grey literature. This was because these abstracts had not undergone peer review. They might have contained preliminary results and did not include final analyses, which limited the researcher's ability to assess their relevance adequately
- Studies involved only adults (i.e., older than 20 years old)
- The researchers had studied a treatment other than orthopaedic surgery to the lower limbs (e.g., upper limb surgery, physiotherapy)
- The papers reported surgery that had been performed only for hip dysplasia

7.2.3 Step three: Study selection process

The collected research was exported into the reference manager, EndNote X8.2, in which duplicated studies were removed. A two-stage screening process was then undertaken. Firstly, the titles and abstracts of studies identified by the search strategy were assessed against the inclusion criteria. Second, a full-text screening was undertaken of all studies that had been identified during the first step.

To reduce the chance that relevant studies might be missed, this process was performed twice. Titles, abstracts and selected full-text articles were independently assessed by two reviewers (HA and NW). HA screened the titles and abstracts to determine eligibility for in-depth examination, and the second reviewer (NW) screened the titles and abstracts of a random list of identified studies. A sample, rather than the complete set, was selected due to resource constraints. HA screened and reread the eligible full texts to double-check the eligibility of each study, and NW was asked to screen all identified studies

from stage one. Discrepancies between the two reviewers were rare and were mostly due to differences in terminology. Disagreements were resolved by discussion to reach a consensus. The third reviewer (TT) was consulted if no consensus was reached.

A PRISMA flow chart was used to identify all search processes that had been used within the final included studies. No studies were excluded based on a risk-of-bias assessment or a judgement of methodological quality, as the purpose of the current study was to identify outcome domains and outcome measures rather than to examine the quality of the data or the treatment effects.

7.2.4 Step four: Data extraction

A pre-designed, standardised data extraction template was developed on Microsoft Excel in order to systematically document essential data (Table 34).

Table 34 Data extraction element

Type	Element
Study data	Authors and year of publication, study title, aim/purpose of the study, the country in which the study had been conducted, and study design
Participants data	Number of participants; age range; gender ratio; GMFCS level; and surgery type
Outcomes and outcome measures	What had been assessed; and the domain of health according to the ICF-CY framework, when these outcomes had been assessed, and the outcomes measures that had been used.

Outcome measures were only selected if at least one published study reported their psychometric properties. This was verified by checking the reference lists of eligible studies and by searching the bibliographical PubMed database for each outcome measure. If the outcome measures' psychometric properties had not been evaluated previously, these were excluded from further review.

Data were extracted by two independent reviewers (HA and NW). Agreement was sought between reviewers and any modifications to extraction were suitably noted. There were no concerns regarding the interpretation of extracted outcomes that had been assessed, health domains or outcome measures.

7.2.5 Step five: Analysis

A descriptive summary was produced that contained each study's characteristics. The breadth of content of each of the identified outcomes and outcome measures was classified with reference to the ICF-CY

domains. Several outcomes that were reported in the literature were suited to the additional domains of adverse effects and harm outcomes, which was a finding that was consistent with previous recommendations (142).

Data were then summarised by use of descriptive analysis, which considered the range of different outcomes and the frequencies and proportions of each reported outcome domain and outcome measures. The patterns of outcome reporting over time were explored overall and within domains.

7.3 Results

The results presented in this chapter were focused mainly on the surgical outcomes of interest, while the results related to the outcome measures are described in Chapter 11.

7.3.1 Selection of sources of evidence

The literature search identified 5,754 studies and screening of the references lists yielded an additional 29 studies. All potential studies (n= 5,783) were imported into Endnote X8, which led to the identification of 2,241 duplicates that were removed. A total of 3,542 titles/abstracts were screened; 3,378 studies were excluded, and 164 full-texts were screened. Of these, 120 studies were excluded. The majority of these excluded studies (68) were conference abstracts; 19 studied non-ambulatory CP; 11 studied adults with CP; 11 were related to hip dysplasia surgery for severe, non-ambulant CP; one study considered other disabilities; data were unavailable in one study; one study did not report outcome measures; and one was a validation study. Thus, the review comprised 44 studies on the effectiveness of lower limb orthopaedic surgery. The selection process details are outlined in Figure 12. Full references for the included studies are provided Appendix 6.

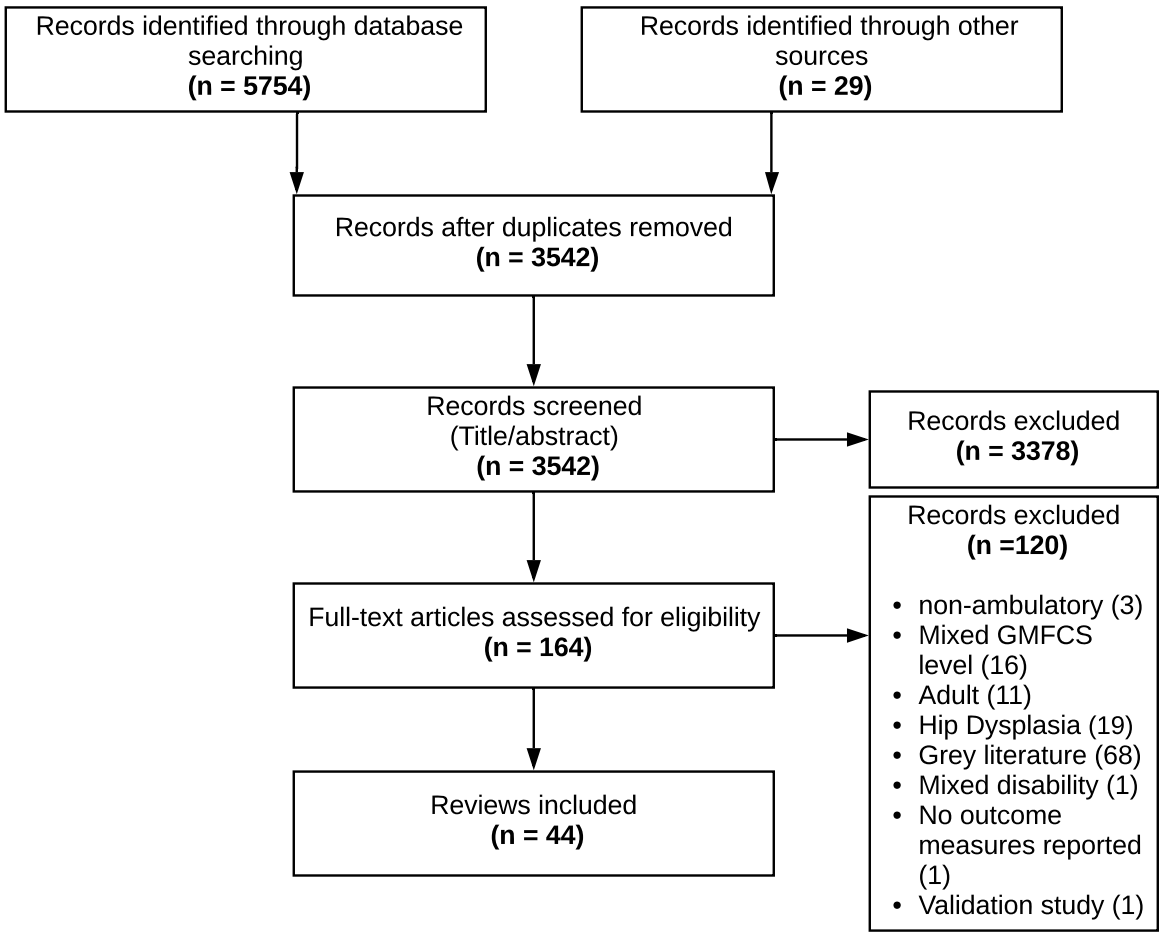


Figure 12 PRISMA flow chart

7.3.2 Characteristics of sources of evidence

Only two (5%) of the 44 eligible papers were RCTs; the remaining 42 articles (95%) were retrospective studies. The 44 studies addressed several orthopaedic surgical procedures in the lower limb, including bony, soft-tissue surgery, and combinations of bony and soft-tissue surgery in the form of SEMLS. Sample sizes ranged from 6 to 314 (median= 34). The majority of studies reported the GMFCS levels (37 out of 44 papers, 84%).

7.3.3 Outcome Domains

There were 184 reported outcomes, which described 40 different outcome domains (Table 35). Outcomes were grouped into the ICF-CY domains body structure and function and activity and participation. Outcome groups were expanded to include domains that related to adverse effects and harm.

All studies identified reported outcomes that related to the impairment of body function and structures. Twelve outcomes that reflected body function and structure impairment domains represented a 48%

usage rate of the total reported outcomes. The most common outcome was 'gait pathology and pattern', which appeared in 37 of the 44 studies (84%), followed by 'joint mobility' (n=20, 45% of the studies), 'muscle tightness' (n=7, 16%), 'muscle strength' (n=6, 14%) and 'pain level' (n=4, 9%).

Only seven studies (16%) reported outcome domains related to the activity and participation level of the ICF-CY framework. Nineteen outcomes related to activity and participation were identified, making up 37% of the frequently of use of the total outcomes reported. Walking-related outcomes were the most frequently mentioned and were represented within five studies (11%). Other less frequently reported outcomes included: 'engaging in sports activity', 'self-care', 'standing and sitting ability' and 'activity engagement level' (Table 35).

Only 30% of the studies (n=13) reported adverse events that had followed lower limb surgery. The most commonly reported adverse event, reported in seven studies (16%), was infection, followed by the need for recurrent surgery, which was reported in five studies (11%). Other less frequently reported adverse events included: surgery failures such as implant failure and non-union, fracture, overcorrected surgery, complex regional pain syndrome and depression.

Table 35 Outcome domains across 44 studies

Outcome Domain	Identified papers (n= 44)		Reference
	Number of papers	Percentage (%)	
Body Function and Structure			
Gait pathology/pattern	37	84%	(164-168, 383-414)
Gait appearance	2	5%	(389, 415)
Joint mobility	20	45%	(164, 165, 386, 387, 390-392, 394, 397-399, 402, 404, 405, 407-409, 416-418)
Joint alignment	5	11%	(385, 394, 417, 419, 420)
Lower limb alignment	2	5%	(164, 389)
Muscle power/strength	6	14%	(165, 386, 387, 397, 398, 421)
Muscle tightness	7	16%	(165, 386, 392, 397, 398, 405, 417)
Energy drive movement	2	5%	(166, 400)
Ankle structure	1	2%	(166)
Knee structure	1	2%	(399)
Pain level	4	9%	(164-166, 415)
Fatigue	2	5%	(164, 415)

Activity and Participation			
Changing body position	5	11%	(165, 166, 415, 417, 421)
Mobility level	5	11%	(164-166, 415, 421)
Game/sport	3	7%	(165, 166, 415)
Outdoor activity	4	9%	(164-166, 415)
Jump/run	5	11%	(165, 166, 387, 415, 417, 421)
Walking ability	6	14%	(165, 166, 387, 415, 421)
Walking in uneven surface	6	14%	(165, 166, 387, 415, 421)
Walking long distance	3	7%	(165, 166, 415)
Using assistive device	4	9%	(165, 166, 415, 421)
Stepping	1	2%	(417)
Using stair	4	9%	(387, 415, 417, 421)
Ride a bike	4	9%	(165, 166, 387, 415)
Sitting in all forms	2	5%	(165, 415)
Standing in all forms	3	7%	(164, 165, 415)
Using transportation	3	7%	(165, 166, 415)
School activity engagement	3	7%	(165, 166, 415)
Self-care	4	9%	(165, 166, 415, 421)
Mobility in home	1	2%	(421)
Social satisfaction	2	5%	(164, 166)
Adverse events and harm			
Recurrent surgery	5	11%	(385, 395, 402, 407, 410)
Surgery failure	2	5%	(396, 407)
Infection	7	16%	(385, 395, 396, 398, 407, 410, 416)
Fracture	2	5%	(407, 416)
Neurovascular symptoms	4	9%	(395, 398, 416, 418)
Healing process	3	7%	(389, 398, 416)
Complex Regional Pain Syndrome	1	2%	(389)
Overcorrection	2	5%	(167, 404)
Depression	1	2%	(164)
Total	184		

7.3.4 Timing of assessment

The timing of the outcome assessment ranged from <12 months to more than 10 years after surgery (Appendix 6). Short-term assessment (within one year) was reported in 16 studies (36%), mid-term

assessment (after three to five years) in nine studies (20%) and long-term assessment (more than ten years) was only reported in three studies (7%). There was also variability in the follow-up point that was used in some studies (n=16, 36.5%); for example, Bittman et al. (419) assessed outcomes at a range of time points between 6 to 36 months after surgery.

All assessments of body structure and function were reported at different follow-up time points; for example, 3D gait analysis to assess gait patterns was used at a range of time points from <1 year up to 10 years post-surgery. Of the studies that reported activity and participation outcomes (n=7), the majority were performed at 6-12 months after surgery (n=5, 71.4% of the studies that reported this outcome) and/or after two to three years (n=3, 42.8%).

7.4 Discussion

This updated review considered additional studies to those that had been considered in the first review by Wilson et al., which was conducted in 2016. The additional studies contained clinical information on the outcome domains that were reported following lower limb orthopaedic surgery in CP. The review identified 44 studies, which suggested that research in this field continues to expand. The updated review provided novel information regarding what was measured, based on the ICF-CY taxonomy. Despite the relatively short period (2016- July 2019), the studies reported measurement of 40 different outcomes and no single outcome was reported in all the identified studies.

The majority of outcome domains that were identified mainly addressed the ICF-CY domains of impairment and functional levels, which was similar to those of the previous scoping reviews (6, 191). The observation may reflect the natural progression of CP in children; which is characterised by deterioration of posture and gait due to musculoskeletal pathologies, such as bone deformities and joint contractures. Therefore, health professionals might focus on the improvement of children's gait outcomes by addressing their musculoskeletal pathology (3).

As observed in the previous review, there was an imbalance in the choice of the ICF-CY domains that were measured in the identified studies, which were weighted toward body structure and function. This may reflect the focus of orthopaedic interventions, which is to address of impairments to the body and its functions although the other categories of the ICF-CY framework may be essential to represent outcomes that are important to the patient and family. Researchers have recommended that

comprehensive measures of health status include all the components that are identified by the ICF-CY framework (6, 141). A recently developed multidimensional outcome measure, the Gait Outcomes Assessment List (GOAL) (3), assesses function in ambulant children with CP in all the ICF-CY levels. This measure has achieved initial documentation of discriminant and concurrent validity; however, it has not yet been extensively adopted in research that considers the outcomes of lower limb surgery.

The findings of this review highlighted that there were no studies specifically measuring QoL. McGinley et al. (110) reported that "the relationships between orthopaedic deformities, function, and gait and both health-related QoL and QoL are poorly understood and are certainly not linear". The majority of the studies involved assessed short-term outcomes post-surgery, whereas a 12- to 24-month time frame may have been insufficient to measure significant changes in an individual's QoL. The findings of this review were consistent with those of the study by Cuomo et al. (422), which found that multi-level surgery had no impact on children's psychosocial well-being and state-of-happiness outcomes over a 12-month period post-surgery. Further research is needed to investigate the impact of the surgery on children's QoL over a longer period.

The review identified limited reports of adverse events. This sparsity of reporting may have indicated selection bias in reported outcomes within studies (5, 110). Repeated surgeries and side effects (e.g., infection, neurovascular complications) were reported as adverse events of surgery; however, these adverse outcomes accounted for less than 25% of the total outcomes that were identified, and they were reported in only 15 of 44 studies (34%). This imbalanced reporting between the benefits and harms can lead to a poor understanding of the impact of surgery on an individual's health (423). Furthermore, clear knowledge of postoperative complication risk is important since it can both inform surgery choice by clinicians and is considered a key element that the patient must understand before giving consent for surgery (424).

Strengths and limitations

To our knowledge, this was the first scoping review that aimed to underpin the development of a COS for children with CP who are undergoing lower limb orthopaedic surgery. This revised review provided novel evidence, which included: a comprehensive overview of the outcome domains that had been assessed in this field of research; signs of a notable shift in the use of patient-reported outcomes, which is becoming common clinical practice; and signs of the under-reporting of adverse events, which remains a major problem in research and clinical practice. Furthermore, the ICF-CY framework was used to

categorise the outcome domains. This was in line with findings that the use of a common framework to describe content has proven to be advantageous to guide the selection of measures (287).

This review had some limitations. Firstly, as in the original review, it included only peer-reviewed articles that had been published in English. Therefore, it may not have included all relevant outcome domains. Another limitation of this review was the inclusion of all peer-reviewed studies, regardless of their methodological quality.

7.5 Conclusion

A broad range of effective outcome domains (n= 40) was identified from the literature with the focus predominantly on children's body structure and function. The review highlighted the dominance of clinical-based assessment, although there has been an increasing shift in recent years among researchers toward the use of, and awareness of the need to use patient-reported outcomes. It was also notable that few studies had reported adverse event outcomes, which is of concern, given the importance of understanding adverse events in the surgical consenting process.

Next step

This review together with the qualitative synthesis (Chapter 6), were essential in identifying potentially important outcomes following lower limb orthopaedic surgery. The identified outcomes and outcome domains were used, in turn, to stimulate further discussion among the key stakeholder groups in the qualitative study that follows. These identified outcomes guided the development of the topic guide questions for the stakeholder interviews. In particular, outcomes related to quality of life resulted in more in-depth discussion by directly asking stakeholders how important quality of life was to them and how they understood quality of life.

Chapter 8 Qualitative Interviews

Outcomes of importance and factors influencing stakeholders' expectations following lower limb orthopaedic surgery: a qualitative study to inform a Core Outcome Set

This chapter describes qualitative interviews that were conducted with relevant stakeholders, which sought to support an exploration of lived experiences of CP, the surgical outcomes that mattered to the stakeholders, and the factors that may have influenced their expectations and perceptions of surgical outcomes. This is the third methodological strand to the research in this thesis. The outcomes identified in this chapter will be carried through to a consensus process to endorse the outcomes to be included in the final COS. This chapter was adopted and extracted from published papers (227, 425).

8.1 Background

In the context of orthopaedic surgery to treat CP, few studies have considered children's and their parents' perspectives of lower limb surgery (209-211). To improve outcomes of this type of surgery for CP, it is crucial to determine the important outcomes for different stakeholders (212, 286). Additionally, in order to present a complete picture of the relevant outcomes, consideration must be given to contextual factors that may influence an individual's expectations and may or may not produce the desired effects of lower limb orthopaedic surgery.

As discussed in the literature review, a guiding principle in the development of a COS is that, in order to maximise the relevance and impact of future research, outcomes must reflect the views and priorities of all key stakeholders, including patients, and their representatives and healthcare professionals (3, 203). The use of qualitative methods can help to ensure that all potentially important outcomes have been identified prior to the consensus process, and that no important outcomes have been missed. Qualitative research also gives voice to individuals and develops rich insight into experiences and views which are not attainable using quantitative methods such as surveys (426, 427).

A qualitative study has been undertaken previously to help to develop priority-based PROMs although few qualitative studies were developed regarding lower limb orthopaedic surgery for CP. For example, Narayanan et al. (428) conducted a qualitative study in 2011 to determine the priorities of children with CP and their parents in order to develop a GOAL questionnaire. Twenty-two children (15 males; 7

females, aged 5-11 years) and their parents were interviewed. The study generated 49 items and five key themes: independence and activities of daily living (ADL); gait function and efficiency; gait appearance/self-esteem; comfort and endurance; and physical activities/sports and recreation. However, the study was published as an abstract only and contained very limited details of the research context.

The QES in Chapter 6 only identified six studies that have specifically investigated the experiences of stakeholders in relation to lower limb orthopaedic surgery for CP. The QES reported in Chapter 6 showed a wide-ranging impact of surgery on issues as diverse as physical, social and emotional aspects, yet none of the studies included sought to understand, or fully explore, what a good outcome post-surgery looked like, or which outcomes really matter to children, their parents and health professionals. This information was required to inform the development of a COS in this field. Moreover, there has been a lack of research specifically focused on the identification of factors that contribute to stakeholders' expectations of surgical outcomes [9].

Qualitative research can help to minimise the number of rounds necessary in the next stage, a Delphi process, by enabling omission of the open 'blank' round, which aims to collect qualitative feedback from the participants in a first-round survey. Minimising the number of rounds can improve the retention of the Delphi participants across rounds.

A methodological review paper (426) summarised researchers' experiences of using qualitative methods in the pre-Delphi stage for three different COSs. It showed that qualitative research can aid identification of outcomes that are important to stakeholders, help with prioritisation of outcomes, determine the scope of outcomes, identify the best language for use in Delphi surveys, and inform comparisons between stakeholder data and data from other sources such as systematic reviews.

Therefore, this qualitative study aimed to:

1. identify important outcome domains from each stakeholder's group,
2. explore the lived experiences of young people with CP and the impact of CP on their lives, and on the lives of their parents, and
3. explore factors that may affects participants' expectations of surgery.

Study Question

The following questions guided this qualitative study: what outcome domains are considered important by ambulant children and young people with CP, their parents and health professionals after lower limb orthopaedic surgery? What factors are considered important by all stakeholder groups to impact their expectations?

8.2 Methods

8.2.1 Research Ethics Committee

Ethical approval for this study was granted in the UK by the South Central - Oxford Research Ethics Committee, under reference 19/SC/0357.

8.2.2 Interview development

Two steps were undertaken to inform the planning of interviews with stakeholders. Firstly, members of the patient and public involvement (PPI) group were consulted. Secondly, a qualitative protocol was developed collaboratively with the project team.

Patient and Public Involvement

Meetings were arranged with PPI representatives to explore and improve the study design. An explanation of the overview and purpose of the project was provided to the PPI representatives and specific advice was sought about several concerns. These were: the best way to approach potential participants; the interview process; the appropriateness of the questions in the interview topic guide; and the wording and content of information documents.

After this discussion, it was agreed that the interviews would focus on understanding children's and parents' experiences of living with the condition, and in particular, their experience peri- and post-surgery, and participants' expectations of surgery.

Representatives suggested that participants should choose the interview location, which could be either their home or hospital based. They felt that one-to-one interviews would offer participants privacy as the discussions would involve personal experiences that might be difficult for many to talk about, particularly in public. Open discussion might also be difficult to hear in a public and group setting. They also suggested amendments to information documents to make them more concise and focused.

Consultation with Research Team

Consultation within the research team was focused on the maintenance of the quality of the research and on ensuring systematic, rigorous research reporting. The interview protocol, consent forms, information sheets and topic guide were reviewed and discussed within the research team.

The Consolidated Criteria for Reporting Qualitative Studies (COREQ) (329) were used to guide the report, as recommended by the Enhancing the QUAlity and Transparency Of the health Research (EQUATOR) network.

8.2.3 Study Design

Semi-structured interviews and focus groups were considered as potential methods of data collection to explore and identify outcomes of importance to stakeholders. One-to-one interviews can provide an in-depth understanding of individuals' experiences and enable participants to control the discussion and ensure that the important points are covered (429). Interviews also offer the benefits of location and time flexibility, causing minimum inconvenience to participants. They enable the participant to talk in detail for as long as they wish about a topic, which may not be possible in a large group setting. Interview settings can be chosen to help the participants to relax and to increase openness in a comfortable environment (430). In a one-to-one interview setting, emotional upset can be resolved, and any embarrassment minimised.

In contrast, focus groups provide the benefit of bringing together larger numbers of people to give their views, at low cost and within a short period (431). Focus groups also produce additive data that represents a range of views and can lead to the development of ideas, which may not occur in individual interviews. However, participants might not feel comfortable talking about personal experiences in a group, the members of which they are unfamiliar with, there would be limited confidentiality in a group. In addition, children with CP are heterogeneous and their experiences of surgery would probably differ, which might cause awkwardness or concerns. For example, a child may have experienced deterioration following surgery, and their discussion of it could make other children in the group worry about planned surgery. Finally, it was also taken into account that the project PPI representatives favoured one-to-one interviews, which could be organised at the convenience and setting of the participants' choice.

In the context of the current study, qualitative, semi-structured interviews were chosen as the data collection method because it was most suited to the objectives, given that the topic had not been explored to date and hence individual interviews would provide greater insight than could probably be gained from focus group discussions.

8.2.4 Participant Eligibility

Inclusion and exclusion criteria for each stakeholder group are provided in Table 36.

Table 36 Participants' inclusion and exclusion criteria

Inclusion Criteria (children, young people, and parents)
- Diagnosed with any type of cerebral palsy
- Aged from 8 to 18 years old at the time of the surgery
- Ambulant, within GMFCS level I, II, III.
- Have undergone or are being considered for a lower limb orthopaedic surgical intervention
- Parent of a child or young person meeting the inclusion criteria above
Inclusion Criteria (health professionals)
- Healthcare scientists/researchers (expert in the fields of clinical research for CP, more specifically in orthopaedics)
- Clinicians, who have at least 2 years of clinical experience in CP: orthopaedic surgeons, paediatrics physiotherapists, occupational therapists, paediatrics nurses, orthotists
- Clinical scientists
Exclusion Criteria
- Non-English speakers
- Children or young adults who are cognitively impaired and unable to self-consent to participate in the study
- Children or young adults with communication disorders

8.2.5 Sample strategy

Participants were sampled to achieve diversity in terms of certain clinical and demographic characteristics such as the children's ages, GMFCS levels, surgery type and time (years) since surgery, and the backgrounds and length of experience of health professionals. These characteristics were likely to have influenced their experiences, perceptions, and priorities. Therefore, participants were chosen through a purposive sampling strategy (432).

In purposive sampling, participants are selected because they meet the criteria that are anticipated by the researcher to be relevant to the research question. In this study, this process was facilitated by production of a sampling matrix. Initially, participant characteristics were recorded against this matrix,

and subsequent attempts were made to identify participants with characteristics that had not been identified in the participant sample in order to fill the gaps.

Eligible children were identified by the local care team, who reviewed case notes in order to collect demographic and clinical data. The children's ages, genders, mobility levels and the period that had elapsed since surgery were recorded. As it was likely that ethnicity would have affected decision-making and how treatment and recovery were negotiated (433), significant efforts were made to recruit participants from a range of ethnic backgrounds. Despite these efforts, no participants from minority ethnic groups were available in the study time frame.

8.2.6 Sample size

Unlike quantitative research, in which power calculations are used to decide on an appropriate sample size, qualitative research does not have equivalent guidelines that can be used to establish how many participants should be recruited. Qualitative studies employ smaller samples of participants than quantitative research because their aim is not to generalise the information, but rather to collect data to explore and gain an understanding of the phenomena under study (434, 435). The sample size would be set to achieve data saturation (whereby increasing the sample size no longer contributes to further insight) (434, 436-438).

Guest et al. (439) reported that data saturation was likely to be achieved with 12 interviews and that new data were unlikely to emerge from a participant group larger than this number. Gareth et al. (440) recommended a sample size of at least six in studies that employed the interview method in order to capture patterned meaning, while Joffe (441) suggested a sample size of 32-80 participants.

As there is no consensus regarding the ideal number of participants for qualitative research and the number of participants that are recruited for such studies varies widely, the research team agreed that the sample size should be sufficient to allow an in-depth analysis. The aim was that the data collection should continue until data saturation had been achieved. Within the current study, the main interviewer (HA) and a member of the research team (FT) agreed on the point at which no further interviews would be conducted. The decision was supported by holding regular debriefs with the research team following interviews, and developing of a data matrix that highlighted new themes and areas of importance as they emerged from the data.

Other qualitative studies designed to inform a Delphi process in COS development have shown that the point at which data saturation occurs varies; for example, in the study of the management of otitis media with effusion in children with cleft palate (MOMENT) (442), data saturation was achieved with 30 participants. In the PARTNERS2 study (443), which looked at mental health conditions, saturation was achieved after 14 interviews. A study that investigated fatigue in motor neurone disease, which involved qualitative interviews and a cross-sectional survey (444, 445), reached saturation at 10 interviews.

It was anticipated that 30 participants in this study would enable the researchers to reach a point of data saturation. Thus, the aim was to recruit 10 participants per group (children, parents, and health professionals). The sampling frame served as a guide and was not used to restrict the numbers of participants, especially at the initial stage of interviews, until data saturation was achieved.

8.2.7 Recruitment

The team members at the Oxford University Hospital NHS Foundation Trust are responsible for planning orthopaedic consultations, surgery and rehabilitation for most children and adolescents with CP in the wider region. For this reason, recruitment of participants who fulfilled the selection criteria was realistic and the population were likely to be representative of the South of England area.

Approaches to children and parents

Potential participants who were due to attend for routine follow-up were identified from outpatient clinic lists. Children's case notes were examined by the clinical lead of the paediatric orthopaedics team to screen for eligible children, and they were either approached at the end of their consultation appointment, or by post, to ask if they were interested to take part in the study. A recruitment poster was placed in the outpatient waiting room to invite potential participants who met the criteria to participate in an interview (Appendix 7).

The potential participants were given information leaflets and time to consider the study information before they consented to an interview. The participant information sheet stated the nature and purpose of the study and made it clear that participants could withdraw from the study at any time. It explained that the researcher would pause the audio recorder and the interview could be stopped at any time (for example if a participant became upset or needed a break). It also explained that the interviewees' quotes would be de-identified in any publication by application of ID codes.

Those who were interested and who were approached at the end of their clinical visit were able to discuss the study in more details with the lead researcher (HA), who was available to talk about the study and to answer questions. Other potential participants were sent letters of invitation and information sheets and were asked to contact the researcher to arrange a convenient time to conduct an interview.

Due to the COVID-19 pandemic, the interview method with potential participants was modified to minimise the risk of transmission. After ethical approval had been received, invitations and information sheets were sent by email to potential participants, who were asked to contact the researcher by email to arrange a convenient time to conduct the interview online. This modification in the interview design was applied to the last five interviews with two young adults and three children and their parents.

Children who were under 16 years old were recruited in the presence of their parents. Although separate interviews might offer a more accurate insight into individual perspective than a joint interview (child with his/her parents), the decision to be interviewed alone or together was however left to the child and parent.

Participants were offered the option of the interview being conducted at the hospital or at home. Following COVID-19 restrictions, all participants were interviewed online via MS Teams. Whatever the location or format, the researcher was mindful that the environment for the interview needed to be conducive to concentration - private, quiet and physically comfortable.

Approach to healthcare professionals

Potential healthcare professional participants were members of regional expert multidisciplinary teams in paediatric orthopaedics, or researchers in Oxford, UK. Invitations to take part in the study and information sheets that explained the study aims were sent by email to potential participants through their publicly available professional details. Those who were willing to participate were asked to contact the researcher directly to make interview arrangements.

8.2.8 Data collection

Interview process

At the beginning of each interview, the interviewer (researcher) discussed the study with the participants and answered any questions. It was reiterated to participants that the interview would be audio recorded. The participants were asked to provide some demographic information (the child's

gender and age, the parental relationship to the child, health professionals' expertise and years of experience), and some health information about the child (mobility level, recent surgery, and any co-morbidities).

Consent

After initial explanations and understanding had been confirmed, written consent was obtained from participants. Participants were informed that they could withdraw consent up to the point of data analysis. Written assent was obtained for children under 16 years old alongside their parent's consent to their child's participation in the study.

Interview topic guide

A semi-structured interview format was used, which comprised open-ended questions that defined the area to be explored. Open-ended questions were chosen to enable the interviewer or interviewee to diverge from the questions or prompts and to pursue a chosen idea in-depth (446). This approach enabled the flexibility to explore these issues without having to adhere strictly to a list of questions.

The topic guide was informed by themes that had been identified in the QES (Chapter 6) and scoping review (Chapter 7) and discussed and agreed with the project PPI representatives for appropriateness. Different topic guides were developed for children and young adults, parents, and health professionals (Appendix 8). Some questions were added to probe interesting themes that arose during the interview process and in light of ongoing analysis.

Interviews initially focused on participants' experiences of various aspects of their day-to-day lives, including experiences of treatment related to the lower limbs. Once a better understanding of the context had been developed, participants were asked to reflect on their priorities post-surgery, how their priorities had changed since their treatment and how their priorities differed from those of other stakeholder groups. At the end of each interview, the main points and priorities were summarised. To ensure that nothing important to participants had been missed, they were also asked if they had anything else they wanted to add.

To modify the interview for younger children, a 'talking mat' was used. A talking mat is an interactive resource that uses picture symbols to facilitate communication and to provide a structured framework for open questions. Previous research has been carried out to examine the feasibility of using talking

mats with people who have various conditions such as CP, aphasia, and intellectual and learning disabilities (447-449). The researcher (HA) collaborated with a play therapist to create cards that showed illustrations that would be familiar to children, to represent aspects of health that aligned with the study aim.

A total of 157 cards were compiled as a resource to facilitate discussion about 10 outcome topics: moving about, senses and pain, mental functions, activities, communication, participation, education, feelings, body functions, and QoL. Participants were invited to choose which of the outcome topics they wished to discuss. Subsets of symbol cards were then presented, relating to the specific outcome selected; for example, when a participant selected the illustrated card for 'activities', they were then presented sequentially with cards that related to moving around, self-care and walking. Participants were asked if each of these symbols was 'very important' (thumb up and smiling face), 'somewhat important' (a child shrugging his shoulders) or 'not important' to them (thumb down and sad face).

Children's personal choices of outcome cards were grouped on their talking mat boards, and their interactions were captured using audio recordings and photographs of completed boards.

8.2.9 Data processing and management

Interviews were audio recorded and transcribed verbatim. Transcription was completed by the lead researcher (HA) within a few days of each interview to facilitate recall about each interview. Transcriptions were managed and analysed using Microsoft Word. This facilitated collaboration between researchers, who could embed comments and suggestions in documents.

It is not possible to guarantee total anonymity of data in qualitative research because the unique combination of circumstances and events could be recognised by the interviewee or someone familiar to them. However, steps were taken to protect anonymity: participants were allocated a pseudonym code, and the names of places or people, dates, or any other personally identifiable information (for example, unusual occupations) were deleted from the transcripts. At this point, interviews were deleted from the audio recorder.

All data was held on password-secured computers and encrypted at the University of Oxford offices. All paperwork relating to the project was be stored in a locked filing cabinet to which only the research team had access. Only the direct-care team had access to the participant's data. A designated member

of the research team had access to the encrypted records and transcripts. No individuals outside this group were allowed to access the data. In line with university's policy, data was archived at the University of Oxford for up to five years. After this period, the data will be destroyed.

8.2.10 Data Analysis

A content analysis of the data using the Framework Approach was used (450). Content analysis enables the analyst to identify important outcomes and to develop a comprehensive outcome list. Although this approach employs an a priori framework to code data, and is, therefore, deductive in nature, it also introduces the flexibility to incorporate emergent, inductive, thematic analysis (451), and to develop new themes from the data that do not fit the chosen framework.

In the first part of data analysis, the interviews were read to gain an overview of the data and to identify potentially important concepts. The ICF-CY framework linking rules were used in order to capture the most precise ICF code (**Table 25** ICF linking rules) (305). Each line of text was coded according to its meaning and was linked to an ICF-CY code. Each code was linked to the most precise ICF-CY category and domain. If the content of a code was not explicitly named in the ICF-CY category, the 'not defined' or 'not covered' categories were applied.

The second part of data analysis aimed to capture the data that did not fit the ICF-CY framework, and this data was analysed thematically (451). Thematic analysis was chosen as it provides a structured methodology for inductive analysis of data into key themes. It provides a structured methodology for the identification of key themes within a data set. The data analysis was guided by Braun and Clarke's (451) account of thematic analysis, as their guidelines offered an up-to-date and practice-based conceptualisation of the method. They identified a six-step approach to the performance of a thematic analysis. They noted that there was no requirement to approach the steps linearly as the researcher might need to move back and forth between the stages. Braun and Clarke's steps for thematic analysis are listed in Table 37.

Table 37 Braun and Clarke's steps for thematic analysis

Steps	Description
Step 1: Familiarizing yourself with your data	Focusing on reading and re-reading the data, noting down initial ideas
Step 2: Generating initial codes	Coding interesting features of the data in a systematic fashion across the entire data set, collating data relevant to each code
Step 3: Searching for themes	Collating codes into potential themes, gathering all data relevant to each potential theme
Step 4: Reviewing themes	Checking if the themes work in relation to the coded extracts (Level 1) and the entire data set (Level 2), generating a thematic 'map' of the analysis
Step 5: Defining and naming themes	Ongoing analysis to refine the specifics of each theme, and the overall story the analysis tells, generating clear definitions and names for each theme
Step 6: Producing the report	The final opportunity for analysis. Selection of vivid, completing extract examples, final analysis of selected extracts, relating back of the analysis, the research questions and literature, producing a scholarly report of the analysis

Interviews with health professionals were analysed separately from those with children and/or parents. Firstly, data were analysed within each group for common themes. As the analysis developed, data were compared across the different groups to identify convergences and divergences. Finally, data were compared across the different groups (e.g., child's gender, age, and time since surgery) and analysed for recurrent patterns that indicated how these characteristics shaped participants' priorities.

Ensuring research rigour

To ensure research rigour, three researchers, who were trained in the procedures of coding the transcripts and in use of the ICF-CY classification system, independently coded all transcripts to check for consistency of the analysis. Two researchers categorised all the identified codes into categories and domains/themes. Differences that arose in interpretation among the researchers were resolved through discussion.

8.3 Results

8.3.1 Participants and interviews

A total of 20 interviews were conducted (10 health professionals, eight children and parent dyads, two young adults with CP individually). Participants' demographic data are shown in Table 38 and 39. All professionals were specialists in the treatment of children with CP and lower limb deformities, with experience ranging from three to more than 20 years. The age range of the children was 10 to 17 years.

The severity of the condition, based on GMFCS level, varied from level I (n=1) through level II (n=5) to level III (n=4). Talking mats were used in three interviews. An example of a taking mat is shown in Figure 13. Interviews lasted from 25 to 53 minutes.

Table 38 Characteristics of the sample (health professionals)

Participant	Gender	Health Professional		Length of the interview
		Years of experience	Professions	
HP 1	F	11-20	Paediatric PT	31 min
HP 2	F	≤10	Paediatric PT	44 min
HP 3	F	>20	Paediatric PT	37 min
HP 4	F	11-20	Biomedical engineer	25 min
HP 5	F	>20	Surgeon	34 min
HP 6	M	≤10	Nurse Practitioner	39 min
HP 7	M	>20	Surgeon	39 min
HP 8	F	11-20	Researcher	45 min
HP 9	F	≤10	Researcher	53 min
HP10	M	>20	Surgeon	32 min

HP: Health Professional PT: Physiotherapist

Table 39 Characteristics of the sample (children and parents)

Children and young people							Parents		Length of the interview
Code	Gender	Age (Y)	GMFCS	Previous surgery	Time from surgery	Talking mat	Code	Relation to the child	
CH01	F	14	II	SEMLS	Pre-surgery	Not used	P01	Father	29 min
CH02	F	14	III	SEMLS	3.6 years	Not used	P02	Mother	33 min
CH03	F	17	I	Soft tissue†	Pre-surgery	Not used	P03	Mother	45 min
CH04	M	16	III	SEMLS	Pre-surgery	Not used	P04	Mother	45 min
CH05	M	10	II	SEMLS	Pre-surgery	Used	P05	Mother	46 min
CH06	M	15	II	SEMLS	3 years	Not used	P06	Mother	56 min
CH07	F	17	III	SEMLS	8 years	Not used	P07	Mother	50 min
CH08	M	11	II	SEMLS	5 years	Used	P08	Father	43 min
CH09	M	16	II	SEMLS	2.6 years	Not used	-	-	33 min
CH10	M	15	II	SEMLS	1 year	Used	-	-	53 min

CH: Children P: Parent SEMLS: Single Event Multi-Level Surgery † Hamstrings, Gastrocnemius Lengthening

8.3.2 Identified Outcomes

Thirty-one outcomes were generated from the data, and these were organised into five overarching themes. Two themes were linked to the ICF-CY framework: (1) body function and structure, and (2) activity and participation. There was a separate body of data that did not fit the ICF-CY framework, and these data were coded and analysed into three overarching themes: (3) independence, (4) quality of life, and (5) adverse events. A list of the identified outcomes for each group is summarised in Table 40.

Theme one: Body function and structure

1.1 Alignment and symmetry

Some participants said that increased lower limb alignment and symmetry were important outcomes of the surgery. Participants described how improvements in alignment and symmetry could have positive impacts on subsequent activities such as those that involve mobility, or could reduce complications, such as the potential development of scoliosis due to poor symmetry.

> *"The thing I am looking at first is whether the surgery has physically achieved what it supposed to achieve, which is a pair of legs which are straight with knees and feet facing forward when the child is standing and walking" HP10*
>
> *"As a parent I would say, I would have loved it if they would have operated, and his leg became absolutely straight." P08*
>
> *"The important results were that … my knees were much straighter, in a straightened position … they're important to me in a massive way, because it's so helpful in daily life" CH09*

In contrast, some health professionals were doubtful that correction of joint alignment would lead to functional improvement, but they considered it significant because parents and children felt that symmetry was integral to a positive body image.

> *"If one leg's turning in or something, it won't necessarily be because it impairs them functionally, but they are very conscious that it makes them look different" HP04*

1.2 Flexibility and muscle strength

Participants felt that increased joint mobility and muscle strength were valuable outcomes of surgery. Similarly, they felt it was important to manage muscle spasticity postoperatively. Participants emphasised that these could have an impact on other symptoms, such as pain.

"For the ambulatory kids, they can have discomfort because of their spasticity or because of joint problems. So that would always be my prime goal" HP05

"Hopefully then, by doing that [surgery], the muscles will be able to strengthen, and she'll be able to walk longer distances without needing her chair" P01

"Stretching is quite important because my Achilles tendons just don't have the time and (are) too small to lift me" CH02

1.3 Mental health

Participants described how orthopaedic surgery could negatively affect a child's mental health. There was a sense that it was critical to maintaining the child's mental health postoperatively, and some felt that there was a lack of mental health support after surgery.

"I have seen multi-level surgery induce mental health problems on children with probably an underlying problem and it came up on the surface as a result of the stressful experience in the orthopaedic treatment" HP10

"I think you have to, if possible, try and evaluate your mental well-being before you consider undertaking something like that [surgery]" CH07

1.4 Fatigue

Health professionals and children described reducing levels of 'fatigue' or increasing levels of available 'energy' as significant outcomes. They noted some negative consequences of tiredness on the child's daily activities, social activities, and emotional functioning.

"If I use my energy I can't walk or I can't breathe energy means a lot to me, it's in my body. If I don't have energy, I can't run around, I can't walk. I can't go out and chase my friends around or tickle them or play with them. I need energy so I can do stuff" CH08

Health professionals described the complexity of fatigue and recommended distinguishing *general* fatigue from *muscle* fatigue.

"Muscle fatigue is something that orthopaedic surgery is supposed to improve by reducing the energy cost of walking. So, in theory, when you improve the mechanics of the legs you reduce the energy costs of walking that should reduce fatigue. But general fatigue, the feeling of fatigue, is a much more complex

phenomenon. It is not just dependent on energy cost. There are other parameters. Some of them being psychological" HP10

1.5 Pain

Participants described reduction of pain as an important outcome of surgery since pain could have a negative impact on the child's daily activities, social and family life. There was a sense that pain was not controlled well enough, or measured, and that at times it could be the direct result of surgery.

"Pain affects sleep. That impacts on parents. That impacts on relationships at home. That impacts on the child's involvement with the rehab. It impacts on their participation in school, participation in life, basic life" HP02

"My legs were burning every day and they couldn't do anything about that. So, I had to go through some burning pain" CH09

Theme two: Activity and participation

2.1 Function in life

Participants said that the child's ability to function was an important outcome postoperatively. Although their level of function might not be regarded as 'normal', it should be at a level that enabled them to do important activities.

"Whether they've got sort of a range of 30 degrees or 40 degrees; if they're able to actually function with that, that's what's key" HP03

2.2 Mobility

Participants highlighted the importance of maintaining mobility level after the surgery. Mobility was defined as the ability to sit/stand, transfer, and move around. Although mobility was considered as an important outcome, some health professionals felt that time constraints in clinical practice were a barrier to assessing a child's mobility level postsurgery.

"My main goal is to know that I can step off a thing and on a thing, and even if not stairs, even if it was a little tiny sledge, I know I'll be able to try and do it. That's one of my short-term goals" CH04

"We're not routinely measuring them. It's purely time in what we do" HP01

Maintenance of body balance during mobility and reduction of the incidence of falls were considered important outcomes postoperatively. Both children and parents described a constant fear of falling; they also described a fear of the child being alone in the event of a fall. Participants described how falls could harm the child's confidence in their abilities, and therefore lead to reduced independence.

> *"It's almost like I've got agoraphobia almost, not that I can't leave my house, just I'm scared of walking around an open space where I feel like I might fall over" CH06*
> *"Hopefully it would improve her balance" P01*

Participants considered walking to be an important outcome after lower limb orthopaedic surgery. The value of walking was described as a multidimensional outcome that could mean different things to different people; for example, the ability to walk further or faster than before, or improved walking ability, with or without the use of an assistive device. Health professionals raised concerns about the children's potential to maintain walking improvement into adulthood.

> *"Hoping to maintain walking ability long into the future … What I don't want to do is do a big operation on somebody, improve their walking for two or three years, and for that then to decrease again when they're adults" HP05*

Some participants highlighted the importance of being able to walk with improved leg alignment. As such, measuring the quality of the gait would be significant. Health professionals' emphasis was on measuring gait parameters objectively, although some noted that the outcome measures available to evaluate gait abnormalities, such as 3DGA, provided only a limited amount of useful information.

> *"Having something objective like gait analysis is quite helpful … that's the marker we use" HP07*
> *"It tells us nothing about how they feel, what difference it's made for them" HP02*

Others highlighted the importance of improving endurance and being able to walk further than they could before the surgery, either with or without assistive devices. However, health professionals emphasised that they would not expect surgery to reduce the need for assistive walking devices. Some highlighted situations in which children's and parents' expectations of walking capacity postoperatively were not aligned.

"If I think of walking there's aspects of walking that matter for health and there's aspects of how fast you can move or how safe your balance, how you look. And maybe the effort of getting around. And I think they're not always the same" HP08

"(the) key focus is to get mechanically their walking as good as possible, which mostly means making their pattern as close to normal as possible" HP04

"I would like to walk longer distances without getting tired" CH01

There were differences between the views of children and their parents, mainly related to the use of assistive devices. In contrast, some participants felt that using an assistive device would increase independence. In an example of this phenomenon, one young adult and her mother described her striving toward walking independently as:

"I haven't had someone say, "Well, you won't be able to walk independently," but I've also had the physiotherapist say, "Oh. Well, probably, our goal for you is to be walking with sticks." And I said, "Well, I don't really want to walk with sticks, so it's not a goal I want to go for." Because, if I'm working so hard, and then that is as good as I get" CH07

"Your goal would be to walk unaided. But, of course, that has to be tempered with a big dose of realism. And I think for [child's name] to go from where she was to being able to walk completely unaided would've been unrealistic" P07

2.3 Participation

Participants described the child's participation and involvement in everyday activities and social life as a key outcome. They described several aspects of participation within the home, the school, or the community. Social life was considered as an opportunity to be involved in recreation and leisure activities and a key aspect of social inclusion. Participants emphasised the fundamental role of participation in sport, and related this to its impact on the child's physical health.

"(I) would probably measure participation. So, whether it's school, if they're going to things with the family, sort of social schooling and family" HP08

"I obviously want to be able to have a healthy relationship with other people, I don't want to be that one guy who has got no friends" CH06

"It's to help [child's name] sort of be able to do more, to be more active, to be more sporty, to enjoy her sport and dance, to enable her to do more things" P01

Theme three: Independence

Being independent in daily life activities, self-care and walking was considered to be an important postoperative outcome. Some felt that it was important to develop independence as children grew up and described struggles to achieve independence.

> *"Independence is a big thing because, before my third year, a lot of people, especially my family, had to rely on helping me every day. That would involve helping me walk, helping me change my clothes because I couldn't, it was very difficult at that time because I couldn't stand up properly" CH09*
> *"She's fully supported by us basically all the time. That's not typical of a 17-year-old … to be able to just undertake more tasks independently and not need that constant supervision" P03*

Moreover, health professionals and children described how challenges of the surgical regime, such as rehabilitation, might threaten personal autonomy and subsequently have an impact on quality of life.

> *"I think it's come up in the sense of autonomy. It's not independence. Autonomy comes up a lot, which is being in control. So, deciding you're going to do something. I think it's probably that's how I've come across that a lot all the time" HP08*
> *"A teenager ultimately wants to be independent and start to push away from their parents a little bit. And it's how we can maximise their independence" HP03*

Theme four: Quality of Life

Global quality of life (QoL) was described as an important outcome by participants although they highlighted that the concept of QoL was complex and that it might incorporate multiple ICF categories. Therefore, for health professionals and parents, making judgements about somebody else's QoL was subjective.

> *"It's a very complex concept and in a child with cerebral palsy it becomes even more complicated because the physical disability may or may not affect quality of life in a different way in different children" HP10*
> *"The number one priority for quality of life is good health and to be pain-free, so you can enjoy" P06*

Participants said that their child's emotional well-being was a main component of QoL and that this was an important outcome. Participants described how feelings and emotions could contribute to QoL. Children could experience stress, fear, anxiety, and depression after surgery. Some described the

exhaustion that followed major surgery as crucial factor in QoL that harmed the child's and family's emotional well-being.

> *"I would measure a kind of one mood construct. So just about how they're feeling with regards to anxiety, depression" HP08*

Similarly, improved aesthetics were reported to be to be an important postoperative outcome, which would increase the child's self-esteem. Some children made a negative comparison between their own bodies and activities and those of their siblings and friends.

> *"Aesthetics is important. That's about self-esteem, and that is hugely important" HP05*

Children also experienced low self-esteem if they had lost their path and passion in life due to the condition. A 17-year-old young adult and his mother spoke about lost motivation and low self-esteem:

> *"The quality of life is very important because there are a lot of things in life that I'm trying to accomplish. A couple of years ago I wanted to become a scientist. I wanted to become the one that used to cure cancer to help people like me. But now, because in my school, they used to give me science classes, but because of the way I am now, I have to come in later which means I'm missing those classes, which means my passion for science went down and because I'm basically stuck in my room for maybe hours a day" CH04*

> *"his self-esteem went down about that because he used to say to me, "What scientist is in a wheelchair?" And I'd say to him that your wheelchair is your legs, so you are no different to anybody else" P04*

Health professionals and parents also linked QoL with components of the ICF-CY framework; for example, they considered QoL to be tied with improvement in the child's physical health and social life, including the ability to get around and to retain a social network.

Theme five: Adverse events

Adverse events following surgery, and being fully informed of potential adverse events, were described as important and integral to the process of balancing the benefits and risks of treatment, and of guiding informed consent.

Health professionals reported several complications that should be measured postoperatively, and parents spoke of complications that their children had endured. Examples of potential problems included infection, fractures, wound problems, nerve and blood vessel damage and pressures sores. They also highlighted a greater attention being paid to reporting of surgical complications, both clinically and in research reports.

> *"I think that [adverse events] is an extremely important aspect of the management. It is very much part of the consenting process before surgery. It is something that should always get up on the scales when weighing the pros and cons of surgery" HP10*
> *"She went through a bit of a tough time afterwards because we were constantly coming back to try and sort out the infections of the wounds" P02*

Talking Mat Example

A Talking Mat was used with an 11-year-old child (CH08) to discuss his surgical goals and priorities (see Figure 13). He began by using the main topic symbols, which helped him to understand the various areas in which surgical interventions might be helpful. He identified the topics/ domains that were the main priorities for his surgery as mobility (leisure activities). He expressed an interest in sport, playing at home or outdoors, engagement in school activities with his peers, and the chance to have enough energy for what he wanted to do and not to become fatigued quickly. He said: "If I don't have energy, I can't run around, I can't walk. I can't go out and chase my friends around or tickle them or play with them. I need energy so I can do stuff".

It seemed from his comments that walking without the use of an assistive device was not as much a priority for him as his parents had thought: "I will use it if my legs are really bad, hurting me". He placed 'walking with an aid' in the middle of the mat and explained that he felt that using it was important sometimes. He indicated that self-care was "Fairly easy, I am not really sure about that"; however, he conveyed it as an important outcome in general, but not for his care.

When presented via the talking mat with topics that were based around the ability to maintain body position and aesthetics, he indicated that these were not priorities for him: "I don't really care about the way I look because we're different". Instead, he spoke in detail about important leisure activities, and stated that he was no longer be able to participate in these activities because of his physical limitations.



Figure 13 Talking mat example of an 11-year-old child

8.3.3 Comparison between outcomes identified and outcomes reported in trials

Outcomes from this interview study were compared with those obtained from the scoping review of published studies in this field (Chapter 7) (227). The present study provided 10 unique outcome domains that had not previously been identified or used in the literature (Table 40). These included mental health, reducing the incidence of falls, improving balance, daily life function, participation in a home context and achieving independence in daily life. None of the outcome domains that were identified under the 'QoL' heading had previously been reported or used in this field of the literature. Furthermore, the assessment of the impact of pressure sores was deemed important as part of the surgical adverse events that had not previously been reported in the literature.

Table 40 Desired outcomes for each stakeholder group and those identified from the scoping review

Outcomes identified	Healthcare professionals	Children and families	Scoping Review (Chapter 7)
Body Function and Structure			
Mental Health	•	•	
Gait pathology	•		•
Joint mobility	•	•	•
Lower limb alignment	•	•	•
Muscle strength	•	•	•
Muscle tightness	•	•	•
Pain level	•	•	•
Fatigue/Energy drive movement	•	•	•
Activity and Participation			
Activities			
Mobility level	•	•	•
Changing body position	•		•
Using stair	•	•	•
Standing in all forms	•	•	•
Reduce fall incidence	•	•	
Balance	•	•	
Daily life function	•	•	
Self-care	•	•	•
Walking	•	•	•
Participation			
School activity engagement	•	•	•
Social satisfaction	•	•	•
Sport/hobby	•	•	•
Within home	•	•	
Other			
Independence	•	•	
Quality of Life			
Emotional Well-being	•	•	
Aesthetics and self-esteem	•	•	
Parent's burden	•	•	
Adverse events			
Recurrent surgery	•		•
Infection	•	•	•
Fracture	•		•
Neurovascular symptoms	•		•
Healing process	•	•	•
Pressure sores	•		

8.3.4 Factors that influence stakeholders' expectations and perceptions

Content analysis and linking rules also highlighted 10 environmental and personal factors that could influence stakeholders' outcome expectations. These were organised into factors that could facilitate or inhibit optimal outcomes, and also personal motivators that could influence the outcome.

Theme one: Environmental factors

1.1 Facilitating factors

The participants talked about five environmental factors that facilitated the best outcome. These were primarily related to healthcare services: interdisciplinary collaboration, communication and information resources, holistic care, shared goal setting, and the accessibility of equipment.

Interdisciplinary collaboration: A multi-disciplinary team (MDT) approach was considered an important element of healthcare delivery. However, the approach was not always seen as holistic, and a parent of an 11-year-old child described the MDT as ineffective postoperatively.

> *"If I talk to the physio, the physio will only talk to me about his physical issues. If I talk to, for example, a medic to do with his mental capacity, then he'll only talk to me about his mental capacity, and the two seem never to talk to each other" P08*
>
> *"The surgery shouldn't just be a single intervention and then is about their life rather than just their one surgery. So much more of MDT holistic" HP02*

Participants also highlighted the importance of physiotherapy input as part of the MDT after the operation.

> *"Physiotherapists are better in looking at the child and a multitude of motor function tasks rather than just their level of walking, like most surgeons would focus on" HP10*
>
> *"The physios and doctors would come every day, do stretches every morning, stretch on that machine which stretches my knee, my leg for about, do my exercise for about 45 minutes every morning. And that was the big, important thing in the strategy. So that's how I think it's successful" CH09*

Communication and information resources: Participants valued interaction and good communication between health professionals, children and parents, and felt that this could have a positive impact on the child's experience and expectations.

"Supporting the child's choices and listening to the child and the parents about what is important. They know their child and they know what the greatest impact is" P03

"Trust is a key, to make sure that they trust us and that we listen to them. We can't block their opinion, but we have to be careful, and especially if the kid is a bit voiceless, so the parents are overpowering. We have to make sure that we hear the kid as well" HP09

Health professionals also described the importance of providing information to families regarding what they should expect over the course of surgery and recovery. They emphasised that provision of information should be based on the individual child's and family's needs as there was no 'one-size-fits-all' approach.

"I think pre-op education, that involves everything. A complete package of what to expect" HP02

"I think there's a balance to be had with those discussions but be led by the patient and what they want to know if it was about information" HP03

Although some parents and children reported that they were well-informed about the surgery, others said that they had not received much information about the surgery or its consequences. A mother of a 15-year-old explained the consequences of poor information postoperatively:

"One thing that I found I would have liked to have been warned about is basically after the operation he had really painful muscle spasms and I found that really scary and I didn't know why they were happening, and I started thinking maybe this is something he has developed as part of the operation and it's not going to go away. And he was in extreme pain and I didn't know why. Whereas if, maybe they don't want to put you off the operation but maybe even just after the operation they could tell you this is a really common thing that muscles are going to spasm and it'll be painful and it's just muscles readjusting to the new position, I think I would've felt better" P06

Participants described two strategies in relation to the provision of information that could improve the understanding of children and families regarding realistic surgical outcomes. First, comprehensive information should be provided before the operation regarding what they should expect from the surgery and sufficient time should be allowed for decision-making and questions.

"I think that a rush to decisions is not a good idea. I have had quite regularly families that have come to me and they have read about it, they heard information about the surgery, they are really prepared and

focused for it. My effort is to rein the enthusiasm in and give them more time for discussions to consider everything, to learn more about this from us" HP10

Second, the children and families should be prepared through viewing of a range of videos of postoperative changes. All participants emphasised the need to present best- and worst-case scenarios clearly to facilitate decision-making.

"I want to know the best-case scenario, the worst-case scenario, and what is the most likely scenario. I think, if I had surgery now, I'd have a much better ability to understand and manage my expectations accordingly. So, that's important" CH07

"We try to show them not only videos of children who have had successful surgery, but also videos of children who were not the top of our results. If you only show them good results, they will assume that only good results come out of the surgery. They have to be aware that's not the case" HP10

"Normally if they tell [child's name] how to go about things, and they maybe then watch a video about it all, like the operation if there was an operation that they could see, so then he could understand it better. Whereas, if you talk to him about it, it does go in, but then he's like, 'Mum, I wasn't sure about that. What do they mean by that?' Whereas if he saw the video, or sort of diagrams or whatever, it gets into his head a bit better" P04

Holistic care: Participants said that the current focus of the surgical assessment was on the child's body structure and function, although they noted a shift toward broadening the assessment beyond the child's body to include their activities and participation. A holistic approach after the operation was described as important to help the child to have the best possible health outcomes.

"We're a bit focused on structural outcomes, body structure rather than the participation and activity" HP05

"Trying to look as holistically as possible so that we've got everything covered" HP06

"There was quite a bit of emphasis on I guess straight leg, having my legs straight and keeping them open. Because I had to wear gaiters for a long time to try and keep my legs straighter" CH06

Health professionals also discussed the essential role of a shift towards patient-relevant outcomes. They were aware of the importance of seeing the child as a person in unique circumstances.

"It's their lives. It's their bodies. It's what do they want? It's not what we can do, it's what do they want" HP02

"It shouldn't just be, 'This is the five main things that we do for post-surgery CP,' because it will depend on each child" HP03

A 17-year-old person shared her experiences regarding this shift in thinking:

"Before, I was scared that I was going to be examined, and they were going to tell me, "Well, this seems good. We need to fix this", and they're looking at something else now. So, it's going in the right direction. Most of the conversations we've had have been concerning my psychological wellbeing rather than my physical wellbeing" CH07

Shared goal setting: Health professionals described the importance of setting goals with children and their parents. They described the importance of learning about goals from the child's perspective, and described patients' reported outcomes as integral to clinical practice. There was also a positive attitude towards parents' goals, and some felt that it was important to include parents' points of view with their children's goals.

"Interestingly, my last couple of visits have been mostly about, 'How are you feeling about this?' Before, when I was younger, it was questions were directed to my mom and my dad. And so, it's better now. They want to know my opinion" CH07

"I think the only way to improve our treatment and get a higher percentage of children and families being happy after the operation is by understanding what makes the outcome from their perspective more positive" HP10

Participants felt that the involvement of children and parents in goal setting had to be considered in relation to the age and maturity of the child. They described young children as potentially seeking unrealistic goals, and recognised that a child's goals might change as they grow older.

"It depends on the age of the child. Obviously. I think when they start getting pubertal, early teens, then that may change and is more of a shared decision-making process. But earlier on, I think they're just going to go with what their parents say to a certain extent" HP07

However, health professionals expressed concerns that, although the patient's and family's goals were important, this view should always be combined with clinical outcomes from a clinician's point of view.

> *"The clinical outcomes are important, and they help us guide the surgeries. And how do we do research if we don't have the clinical outcome? But actually, there should be a bigger focus on what it meant for the patient and to be able to pass that on to other patients" HP02*

1.2 Inhibiting factors (barriers)

The participants discussed two inhibiting factors or barriers that could have an impact on patient experiences and expectations of surgical outcomes. These were: mismatch and divergence of expectations; and lack of time and resources.

Mismatch and divergence of expectations: Some participants described a match in outcome expectations amongst children, parents, and professionals; however, others described discrepancies between what was possible and what was hoped for, that might have a negative impact.

> *"Much as you explain that to parents, there's still this element of hope that their child will exceed the expectations that the surgeon has. I think that's human nature" HP05*
>
> *"The levels of expectation may differ but that's because I suppose the levels of reality are different between adult and a child. We understand a bit more than a child as to what you can realistically expect to achieve against what you [child] would ideally like. And they're never going to be the same" P01*
>
> *"There is differences but other than between the health professional with the children, but it's more about between their children" HP02*

However, health professionals expressed deep concerns about the need to be sensitive when negotiating realistic expectations and the impact that this might have. For example, children and parents might expect full recovery more quickly after surgery than is likely, and it could also be difficult for children to conceptualise the period of recovery.

> *"Some patients' expectations are, I'm going to be able to walk, I'm going to be able to run. It's going to be fantastic. And that's true, but their time frames are messed up because they're children, and of course as an adult a year is a year and as a child a year is forever. Like it's the longest time in the world" HP06*
>
> *"Immediately post-surgery, she didn't feel the goal had been met. She'd obviously thought this would happen very quickly after surgery despite us trying to meet those expectations" HP03*

Health professionals described their experiences of unrealistic expectations from children and parents who thought that they would be able to return to a 'healthy normal life' without limitation. Parents described 'return to normality' as a benchmark of surgical success.

> *"I think expectation around goals can be a little bit more around really normalising their child" HP03*
> *"She wants to feel more normal within an age-appropriate range" P03*
> *"I want to get to live my life more like, if you will, a normal person" CH06*

In contrast, health professionals described the importance of explaining to the child and the parents that surgery would not return the child to 'normal'.

> *"I would not promise normality to anyone really because orthopaedic surgery improves the mechanics of the lower limbs and helps to extend with walking and motor functions if it works well, but it doesn't restore normality. What I would normally tell children and families is that the problem arises from the brain and the operation is done on the legs. It's like we are not treating the problem where it is, but we're treating some of its mechanical expressions" HP10*

Lack of time and resources: Health professionals highlighted the impact of time constraints in the clinic that prevented them from being able to measure desired outcomes.

> *"If you want to measure a lot of these things, it will take time. And also, depending on how you measure them it can take even longer" HP07*

Participants also described uncertainty about the amount of healthcare service input compared with optimal practice. Parents commented that the lack of continuous health services formed a barrier to the best outcome following surgery.

> *"They've had to reduce down the number of times that you can see a child. And we know that the rehab of post this type of surgery is so crucial to the outcome" HP03*
> *"Occupational therapy seems to be overburdened and I haven't been very impressed with the local OT [occupational therapist]. We've not had much help there. And I need to chase it up somehow. Yeah, that's the major thing that I would like with OT, because sometimes you don't know how to teach them things*

like putting his socks on or things like that because there are different ways of doing it that you wouldn't see" P06

Parents described the importance and impact of having rapid access to equipment to improve surgical outcomes.

"You need to assess the situation they're in. Do you have the facilities at home to aid with the recovery? Will you need to move a bed downstairs, a commode, to make sure you've got everything you need?" P01
"Equipment-wise at home, we've gone from having paediatric up-down chairs that could be moved around to a stairlift to a bath hoist, and all that is gone. It's a normal house now" P02

Theme two: Personal factors

2.1 Motivational factors

The interviews identified personal motivators that could influence the outcome: family encouragement, patients' self-determination, and previous experiences.

Family encouragement: Although participants described parents' overwhelming responsibility, and the physical load that was involved in caring for a child with CP, they highlighted that good support and interaction between parent and child could encourage and motivate.

"I just want to go back to work, I do. But I can't at the moment. So, I'm on my own looking after [child's name]" P03
"I don't think you can put in the amount of work that's needed after the surgery to get the best benefit. Because it was like 24-hour care really" P06
"I think the parents take a lot of responsibility to engage their child, to encourage" HP02
"I want my family to support me, and challenge me a bit more, like walk, walk up the step" CH02

Patients' self-determination: Health professionals and children described personal characteristics, such as self-determination, the drive to be the same as everyone else and to be independent, and aesthetic motivations.

"The child's motivation always seems to be, they just want to keep up with what's happening around them and they don't want to be left behind. And I think that's a great motivation" HP02
"I think sometimes motivations are not spoken, and that can be, often, along the aesthetic line" HP05

"Considering how big a deal the operation was, I didn't want to waste it or anything, I didn't really want to end up with [other people] helping me in the end" CH06

Previous experiences: A positive previous experience in hospital was also felt to facilitate the child to keep up with the surgical regime and to stay motivated.

"I've had the surgery; it was very successful. It helped me in many ways. It had a big impact on my life in a good way. I can do a lot of many good things in my life now that I wasn't able to do before and it was just a big success, so I would recommend it to them" CH09

"A lot of their experience in hospital can be based on their previous experiences. Some of the things we do where we've done the right side and then they've come back in to have the left done, you get a lot of anecdotal about how it's made so, for example, some of the foot surgeries that we do, how it's made life so much easier. Their shoes are fitting better, their AFOs [ankle-foot orthoses] are easier to fit. And so, they're then excited about the prospect of having the opposite side done" HP02

2.2 Demotivational factors

The participants identified fear of new environments as a personal demotivator that might have an impact on a child's motivation after the operation.

Fear of new environment: Participants described fear, anxiety, and stress of the unknown or feeling frustrated in a new environment, which might demotivate the child and family pre- and post-surgery.

"A lack of experience means that it is a huge amount of fear of the unknown" HP06

"They might have no experience of hospitals and as such there's the chance that just walking in the hospital doors is terrifying. Whereas, you know, for us walking in the doors, this is every day. I suspect that quite a lot of it is anxiety-led" HP06

To reduce the levels of fear and uncertainty, participants suggested that meeting other families could help to motivate the child toward the surgery and manage their expectations.

"I think meeting with other families who have been through the surgery can be helpful" HP05

"A support network for parents, again, would have been really useful, really useful. I think it would've helped us and it would've helped [child's name], ultimately" P07

Health professionals felt that demotivation could have an impact on the outcome of the surgery.

> *"I have certainly seen poor results in children or families who weren't motivated enough after the surgery to achieve a good result" HP10*

8.4 Discussion

Stakeholders' important outcomes

This study used a qualitative approach to identify 31 outcomes that mattered to children and young people with CP, their parents, and healthcare professionals. These were categorised under five core areas: body function and structure, activity and participation, independence, quality of life and adverse events.

Participants viewed outcomes as interrelated. Some interviews said that one outcome might facilitate another, higher-level outcome, such as life impact. For example, participants described the importance of improving joint flexibility and muscle strength in order to be more active, and this in turn would improve quality of life. This inter-relationship of outcomes has previously been identified and described for children with neurodisability (286). This may indicate the need to consider and identify the degree to which a higher-level outcome reflects clinical and physiological outcomes, and whether clinical and physiological outcomes are the stepping-stones to the achievement of higher-level outcomes and longer-term changes. These findings indicate the need for a comprehensive assessment approach as highlighted by the ICF-CY, to illustrate the interaction between outcomes and their impact.

The findings of this study included ten additional outcomes beyond those that had been identified in a recent scoping literature review (Table 40) (227). In particular, participants considered surgery in relation to the 'whole' person and attached special significance to outcomes that were related to mental health, emotional well-being, independence in activities of daily life, participation and life impacts, rather than outcomes that were aligned to body function and structure, which have dominated the literature. This may demonstrate that current outcome reporting in clinical trials fails to report outcomes that are important and relevant to children and their parents. In contrast, the outcome priorities that were identified in this study, such as achievement of independence, resonate with findings from previous qualitative work with children with neurodisability (284, 286).

A tendency to report clinical outcomes and not life impact is a common feature of published studies. For example, although the impact of surgery on the child's QoL was considered important in this study, it was not reported in any of the 44 studies included in the scoping review (Chapter 7) (227). Although WHO provided a clear definition of QoL as "an individual's perception of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns" (452), there may be reasons why QoL is not routinely reported: first, the complexity of the concept, which encompasses social, emotional, cognitive, physical and functional well-being; and second, uncertainty regarding what should be measured to determine QoL in clinical practice (453). It may also be that improvement in other domains is a key factor towards achievement of a better QoL. The findings of this study support the use of a combination of clinical outcome measures and PROMs, such as the GOAL questionnaire (3), in the evaluation of surgical interventions and clinical trials. Similarly, the findings support the use of qualitative approaches to complement quantitative research findings and to provide detail on surgical impact from the individuals' perspectives.

The findings of this study indicate that there are overlaps in outcomes valued by stakeholders. It seems likely that different stakeholders would emphasise different outcomes, and that healthcare professionals might emphasise outcomes related to their own fields of expertise (for example, surgeons might focus on outcomes related to body structures; nurses might focus on adverse events post-surgery; therapists might focus on activity and participation). Similarly, parents might be concerned about their child's future independence and integration into the community, whereas children and young adults might focus on areas of immediate relevance and their current abilities. This variation in focus has been shown in previous studies (282-284).

The range of identified outcomes indicates the importance of having a multidisciplinary approach and of engaging children and their parents in decision-making. It also highlights the importance of clear, open communication in the decision-making process for surgical interventions and the establishment of treatment goals (454). Incorporation of stakeholders' views about valued outcomes is vital to facilitate a patient-centred approach in health care. This is in line with the aims of generating a list of outcomes that are relevant to all stakeholders, and which includes the voice of children and their parents. This will help to ensure that outcomes of lower limb surgical interventions for CP are not restricted to the treatment of gait pathology and improvement of specific surgical outcomes, but that they include life impact outcomes such as functional, participatory and QoL dimensions of health.

Factors that influence stakeholders' expectations

This chapter reports ten factors that could influence stakeholders' expectations and subsequent perceptions of surgical outcomes. These were organised into four sub-themes: facilitating, inhibiting (barriers), motivational and demotivational factors. These findings add insights to support healthcare system changes to provide optimal care to ambulant children with CP.

The study describes the importance of interdisciplinary support in CP care. The complexity of the condition can make treatment challenging and requires a multidisciplinary approach, particularly when undergoing major surgery. The availability of teams that hold special CP competence to assist in challenging surgical situations in terms of safety and the physiological, psychological, and social impact on the child's pre-and post-surgery care is likely to improve the quality of the CP care. Mineiro et al. (455) emphasised the importance of a multidisciplinary approach when considering the surgical correction of spinal deformities in CP, and noted that this approach improved not only the outcome of surgery but also the patients' and parents' QoL and satisfaction.

The study highlights a shift toward understanding children's and parents' priorities and requirements following lower limb surgery by applying a holistic approach to goal setting. Briffa (456) reported that understanding children's and parents' priorities and goals may have implications for the improvement of compliance with and perception of surgical outcomes. This study highlights potential challenges to a holistic approach due to time constraints, availability of equipment and limited resources such as availability of rehabilitation services. The study indicates that adequate access to rehabilitation services for those with CP who are undergoing surgery is integral to the delivery of good outcomes. This resonates with a previous review by Van Bommel et al. (108).

Beyond multidisciplinary and holistic approaches, the findings support the need to build on communication and provide clear information. It is to be expected that children and families who face surgery may not understand all the information given to them and that they may have unrealistic expectations. These findings confirm the importance of providing clear information and ensuring that patients understand this information prior to surgery. The study findings indicate that parents wish to receive as much information as possible in order to make a fully informed decision about surgery, and that better-informed children and parents might have more realistic expectations of the surgery. These findings are consistent with those of other studies, demonstrating that patients highly value complete information (457-459).

Motivation, such as through family or parent encouragement and the intrinsic reward of achieving valued outcomes, was identified as a facilitator of positive expectations. This, in turn, encourages children to adhere to the surgical regime and postoperative rehabilitation. Bartlett et al. (460) highlighted the importance of personal factors (e.g., the motivation of a child with CP) and family factors in the facilitation of improvement in motor abilities. This resonates with the findings of the study reported here and indicates that it is important to consider contextual factors as key elements in the clinical reasoning and decision-making strategies when the ICF-CY framework is applied to clinical goal setting (461, 462).

Previous experiences of clinical activities, preoperative anxiety and uncertainty of the unknown were considered to play roles in the enhancement or hinderance of child and family motivation and to influence their perceptions of surgical outcomes. Some described orthopaedic surgery as a stressful experience that evoked memories of previous experiences and feelings during different stages of surgical recovery. These phenomena are in accordance with the findings of other studies in paediatric surgery (361, 463). Thus, the provision of educational and realistic information about the potential effects of surgery, and the risks of complications or expected adverse effects, is considered a prerequisite for children and families to be able to make informed decisions about the surgery.

Strengths and limitations

The study identified additional outcomes that had not been identified by a recent literature review of CP surgical trials (426, 464). The coding and analysis were undertaken by a minimum of two experienced researchers.

A potential limitation of this interview study was that participants were stakeholders in one orthopaedic hospital. This hospital is a tertiary referral centre that covers a wide geographical area of South England. There may be outcomes that are relevant to other national and international stakeholders that have not been identified. The interviews were limiting to UK-only participants for pragmatic reasons. However, the next stage (Delphi study) was planned to capture stakeholders' views internationally.

A lack of cultural diversity in the sample is another limitation to be considered. It is important to understand how these factors could impact children' and parents' perspectives of surgical outcomes of interest. Qualitative research does not report statistical findings and does not aim to represent a

particular population; rather, it aims to generate ideas from a purposive sample that will improve understanding of a particular experience. Further research must be done to enhance our knowledge base of the role of sociocultural variables in this field of research.

A further limitation of this study was that most of the children and parents were interviewed together, and this may have had an impact on the responses of either parent or child, which may have been different if they had been interviewed alone. The team considered that the most ethical approach was to give the children the choice to undertake an interview with or without a parent present.

8.5 Conclusion

This study identified 31 outcomes that were important to stakeholders involved with lower limb orthopaedic surgery in CP. Some of these had not been previously identified or used in trials within this research field. It is important that future studies in this field report outcomes that are considered important by children, young people, and their families.

The study also highlights factors (n= 10) that influence outcome expectations and perceptions of lower limb surgery in CP. The facilitators and barriers that were discussed by participants can be supported or minimised by the involvement of person- and family-centred care services, holistic approaches, team-based care provision and provision of adequate information for shared decision-making. If these elements are integrated into healthcare practice, they are likely to enhance the stakeholders' post-operation experiences.

Next steps

The outcomes and factors identified in this chapter (n= 41) were combined with the outcomes identified in the qualitative evidence synthesis (Chapter 6, n= 18) and scoping review (Chapter 7, n= 40) to create a comprehensive outcomes **long-list of 99 outcomes**. The long-list outcomes were used to generate question items for an online Delphi survey as the first step in the consensus process for core outcome set development.

Chapter 9 Delphi Process

Development of a core outcome set for lower limb orthopaedic surgery for children with cerebral palsy: an international multi-stakeholder Delphi study

This chapter presents the results of the Delphi study that was performed to reach a consensus on the outcomes that should be included in a COS for clinical trials of lower limb orthopaedic surgery for ambulant children with CP. This is the fourth methodological strand to the research in this thesis, which brings together the outcomes that were identified in the qualitative evidence synthesis, the scoping review described in chapters six and seven and the qualitative interviews that were described in chapter eight. This chapter was adopted and extracted from a published paper (465).

9.1 Background

As discussed in the introduction to this thesis, variations in reported surgical outcomes challenge translation of evidence into clinical practice, and this results in research waste (256, 466). It is recognised that there is a need to develop a COS for research regarding lower limb orthopaedic surgery in CP, in order to improve the quality of evidence (6, 227, 295). In accordance with the COMET initiative, a key component of a COS development is the elicitation of the views of the stakeholders regarding outcomes of importance. These views should be included as researchers reach a consensus about which outcomes should be included in the COS.

Consensus development is important for researchers and clinicians to make recommendations when there is limited or conflicting evidence from which to draw conclusions (467). Consensus methods are used to define levels of agreement amongst individuals on a controversial issue (467, 468). It is evident that a COS must be applicable to and adopted across international settings and relevant disciplines to enable comparing and contrasting of all research in a topic area. Therefore, consensus methods could be used to gather international expert opinions on COSs.

Thus, this study aimed to establish prioritisation and develop an initial consensus, at an international level, among children, parents and health professionals on a set of core outcome domains for lower limb orthopaedic surgery for ambulant children with CP.

Research question

The research question for this study was: Which outcomes do children and young people with CP, parents of such children and healthcare professionals, think should be included in a COS for lower limb orthopaedic surgery?

9.2 Methods

Three different approaches can be used to achieve consensus (467): two are described as *structured consensus* (the Delphi method and nominal group technique); and one takes the form of *unstructured consensus* (consensus meeting or group discussion). Table 41 summarises the three consensus approaches.

Unstructured consensus methods have a number of limitations, which include the domination of the individuals, pressure on participants to agree; extreme views receiving greater support than they would on an individual level, and the likelihood that complex issues remain unsolved (469). In contrast, a structured consensus is a strong method because it is not conducted face-to-face and therefore its use avoids the problem that more vocal or apparently senior participants may dominate the group discussion and therefore influence others' votes, as is a risk with a patient/clinician cohort (470, 471). Secondly, structure consensus approach such as Delphi surveys enable the achievement of international consensus with input from large and diverse panels without incurrence of additional costs.

Table 41 Characteristics of consensus methods

Consensus approach	Description	Ref
Delphi survey	Multiple rounds of survey with feedback between rounds and the opportunity to alter responses. Usually starts with a qualitative question to generate information and ideas before seeking to develop consensus	(472)
Modified Delphi survey	Starts with consensus development of items informed by various sources, which include literature reviews, interviews, focus groups and panel meetings	(473)
Nominal Group Technique	Face-to-face meeting with five steps: 1. introduction and explanation of the concept, 2. the silent generation of individual ideas, 3. sharing ideas, 4. group discussions and 5. voting and ranking exercises	(474)
Modified Nominal Group Technique	Involves a pre-meeting questionnaire	(475)
Consensus Meeting	Face-to-face meeting of all interested parties with experts. Experts or panels present findings to inform their decision with the opportunity to ask questions	(468)

In the case of our study, a two-stage development consensus process was adopted to prioritise and reach consensus on outcome domains for lower limb orthopaedic surgery. These were: an international modified online Delphi survey; and an international consensus meeting (further details of which are described in Chapter 10. The chosen approach was based on several reasons as described in Table 42.

Table 42 Reasons for chosen consensus methods

Reasons
To maximise the study sample size
International participants could take part
Anonymity and confidentiality could be maintained
The study involved two stakeholder groups, so dominance by those with authority would not influence others' opinions
Low costs
This approach provided the benefits of both an electronically conducted and face-to-face discussion

The modified Delphi process has become a well-established way to reach a consensus among a panel of experts in various healthcare areas. For example, it has been used to capture the views of international experts in the development definitions of PROMS (317); reporting checklists for patient and public involvement (476); consensus development of a definition of neurodisability (477); and the development of Core Outcome Set-STAndards for Reporting (COS-STAR) by COMET initiative (273). Gargon et al. (478) conducted a review of 198 COS studies that considered methods to select the outcomes that should be reported. The results indicated that single or multiple consensus methods should be applied and suggested that a Delphi survey method (or modified version) in combination with a face-to-face meeting could be beneficial. A review in 2018 (479) showed that 85% of COS projects that had been collected on the COMET database used a Delphi survey or modified versions of it. It was evident that combining the benefits of each method increased reliability, consensus and understanding (480, 481).

A detailed study protocol that defines the objectives of the study, the Delphi process and the criteria that were set for participant selection has been published previously (295). This study was approved by Oxford Research Ethics Committee (19/SC/0357). There is no formal guidance for the reporting of e-Delphi studies, so the study followed recommendations by Sinha et al. (482).

9.2.1 Study Design

In this study, a two-round e-Delphi survey was conducted via the online platform Jisc (formerly BOS) to gain consensus on the outcome domains that had been extracted from the qualitative synthesis, scoping review and qualitative research. The Jisc platform was chosen because it was easy-to-use, could provide

an unlimited number of responses and enabled sample dataset extraction. A two-round e-Delphi was chosen instead of a three-round survey because, although the use of more rounds could increase the likelihood that opinions would converge, participation in too many rounds may have resulted in participant fatigue and possibly an increased attrition rate (483).

9.2.2 Survey Development

Five elements were discussed with the research team as part of the development of the survey: the generation of a 'long-to-short' list of outcomes; the scoring system to be used; how consensus would be defined; the survey structure; and how the survey should be piloted.

Generation of a 'long-to-short' list of outcomes

As previously reported, the Delphi survey was informed by the outcomes that were identified by the qualitative evidence synthesis (n= 18), scoping review (n= 40) and interviews with key stakeholders (n= 41) (Chapter 6, Chapter 7 and Chapter 8 respectively). These were used to identify 'long list' outcomes that were potentially important to ambulant children with CP and parent and health professionals following lower limb orthopaedic surgery interventions (227, 295, 307). These three studies identified 99 outcomes across the ICF-CY domains, and the additional domains: quality of life, independence and adverse events.

Face-to-face meetings were held among research teams in Oxford, UK, in preparation for the e-Delphi survey. At these meetings, duplication was removed and outcome grouping. Performance of this process led to the identification of 37 distinct outcomes, which comprised: 26 positive outcomes; six negative side-effects of the surgery; and five composite outcomes that were not related to the intervention, such as availability of information and physiotherapy services.

Keeley et al. (426) and Mathers et al. (427) suggested that a survey should not present just a list of outcomes, as patients and parents might find this difficult to understand. Therefore, it was decided that the outcomes would be displayed as a questionnaire, in the form of a list of eight questions that covered the effects of treatments on outcomes. Each question represented a domain, in which similar or related outcomes could be viewed and rated together. Previous COS work had developed and used this process; for example, a COS for asthma in children by Sinha et al. (484).

Ultimately, the 37 outcomes were combined and arranged into eight domains (Figure 14): pain and fatigue; motor function (symmetry and lower limb alignment); life impact (daily life activities and social life), independence, gait function and appearance; quality of life; adverse events; and factors that affected decision-making regarding surgery. The list of outcomes can be found in Appendix 9

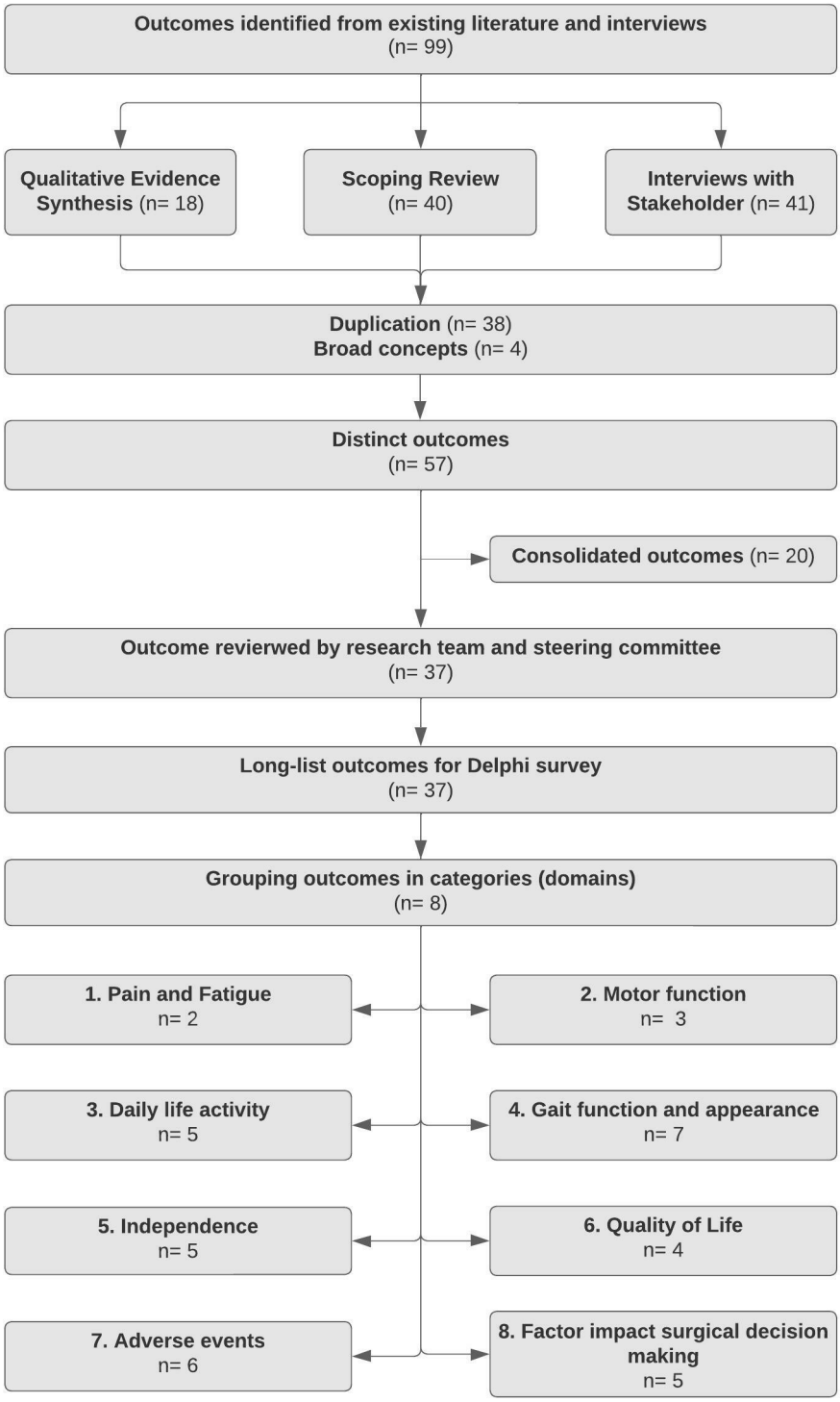


Figure 14 Flow diagram illustrating the pre-Delphi stage of identifying long-list outcomes

Scoring system

Different response scales have been used in previous Delphi studies, through the use of which participants can indicate the importance of the outcomes. These include a Likert scale, which is recommended by the Grading of Recommendations Assessment, Development and Evaluation (GRADE) working group (215, 442, 485). On this scale, the score 1-3 are deemed not important, those that score 4-6 are deemed important but not essential, and those that score 7-9 are deemed essential (485). In other studies, participants have been asked to rank the list of outcomes for their relative importance (486); and in other studies, participants have been asked to divide a total number of, e.g., 100 points between all of the outcomes (487). To ensure that participants considered all the listed outcomes, the use of questions that sought rating was considered to be the most appropriate method for the nature of this research.

The choice of rating scales was likely to impact the final COS because it would influence the number of outcomes that could be made available to the consensus meeting that would follow the Delphi process. It was argued that the use of a scale with too few response options might not allow a respondent to make full use of his or her capacity to discriminate, while the use of a scale with too many options might cause cognitive overloading for participants and contribute to measurement error (488, 489). However, Streiner et al. (490) reported that a nine-point scale would be advantageous over a five-point scale as the former would support the discrimination of importance and reduce the risk of end-aversion bias.

De Meyer et al. (491) compared the use of a three-point and a nine-point scale to rate preliminary outcomes. The authors reported that the use of the nine-point rating scale resulted in the rating of almost twice as many outcomes as important compared with the three-point scale in the first-round of a Delphi survey. In contrast, Remus et al. (492) compared the use of a five-point scale and a nine-point scale, and found that the five-point scale demonstrated greater congruency than the nine-point scale. These outcomes were included in the final pelvic girdle pain COS. The authors stated: “Too many outcomes after each Delphi round is challenging because the goal of this process is to narrow down a larger list into a minimum set and a COS with too many outcomes may not be feasible or may not be ultimately adopted in research and clinical practice” ((492), p2).

De Meyer et al. (491) recommended the use of a nine-point scale over a three-point scale when the aim was to inform the consensus process for a subsequent rating or face-to-face meeting. They emphasised that the three-point scale might be preferred during determination of the final consensus. Nine-point

scale is widely used to measure levels of importance. There are statistical considerations in support of the use of a long scale, which include the ability that such a scale affords to calculate variance in scores (485, 493). Thus, the nine-point GRADE scale was chosen for this study. Figure 15 illustrates the scoring system of a nine-point Likert scale.

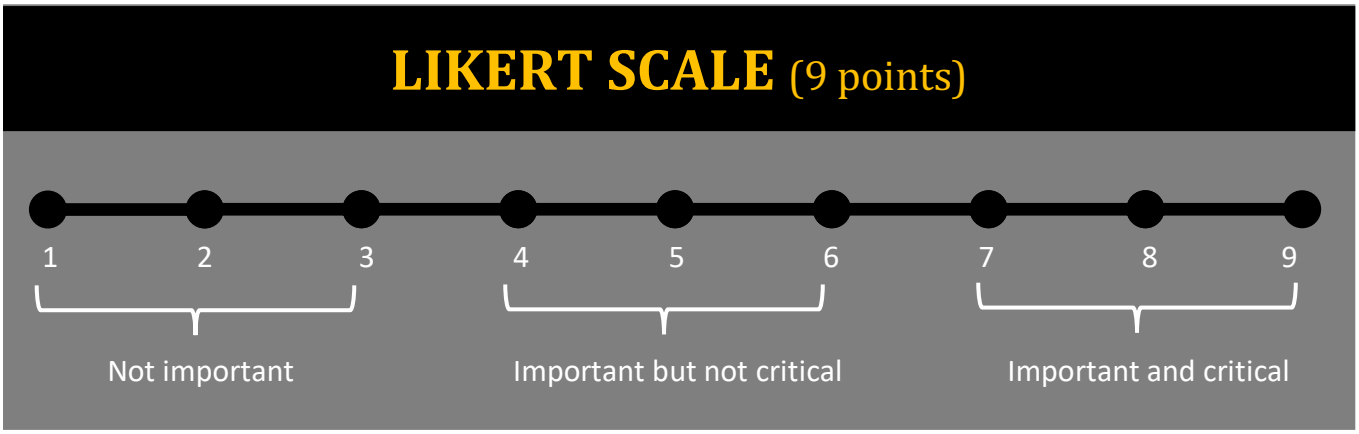


Figure 15 The scoring system that was chosen for outcomes

Defining consensus

There are no agreed methods for the selection of consensus (cut-off) criteria. Different criteria have been used to define consensus in previous COS studies. For example, in a study to define a COS for otitis media with effusion in children with cleft palate, Harman et al. (494) implemented a threshold for inclusion in the core set of 70% of participants scoring outcomes as 7-9 and 15% or less scoring 1-3 to be met by both the clinician and patient panels or 90% or more scoring 7-9 from any single panel.

We took advice from the COMET initiative (10) and decided that consensus to carry an outcome through to the COS would be reached according to the criteria that are illustrated in Table 43.

Table 43 Predefined consensus criteria

Consensus level	Criteria
Consensus in	More than 75% of participants scored its importance as 7 to 9
Consensus out	More than 25% of participants scored its importance as 1 to 3
No consensus	The outcome did not meet these criteria in each panel

For each outcome, it was decided that the number of participants who had scored it and the distribution of scores would be summarised according to panel and stakeholder groups, and each outcome would be classified as ‘consensus in’, ‘consensus out’ or ‘no consensus’. If consensus had not been reached

after round two, or there was major disagreement, a face-to-face meeting was planned with stakeholders to finalise the COS.

Survey structure

Good-practice guidance in questionnaire formatting requires that instructions to participants are clearly visible and easy to read (495). This survey began with a question to discover whether the participants was a child or young adult with CP, parent, healthcare professional or researcher. This was followed by an introduction page, which explained the aim of the research and what the participant should expect from the survey. It ended with a question that sought their consent to take part. Brief demographic questions were asked; for example, if they were a child, their age, time of surgery, their level of mobility, and, in the case of health professionals, their level of expertise. A clear instruction page was provided. It was explained that the survey consisted of two rounds of questions and that they could withdraw from the survey at any point. Finally, the scoring system was illustrated with an example.

Information provided to participants was tailored to the individual groups to enable them to give informed consent and to prepare the participants to be able to score and prioritise outcomes (10). Hence, the introduction and instruction pages were different in terminology according to the stakeholder groups that would complete the survey, to ensure understanding and readability. A child-friendly, animated video hosted on the lead organisation's YouTube channel (496), which provided a visual overview of the project, was presented.

The participants were directed to the survey's main page, on which the outcomes were presented according to their linked domain. Each domain was illustrated on one page. The outcomes were listed in a table and the GRADE scale was presented for each outcome question. The survey also offered participants the opportunity to provide any feedback they wished for each outcome.

Participants were provided with space through an additional open question to provide general feedback or to list any additional outcomes that they considered to be important and missing from the list. A thank-you page was shown after the completion of the survey. A similar structure was taken in round two, with additional information that explained any changes that had been made and how other participants from the same stakeholder group had scored the outcomes. Figure 16 shows how participants accessed the e-Delphi survey.

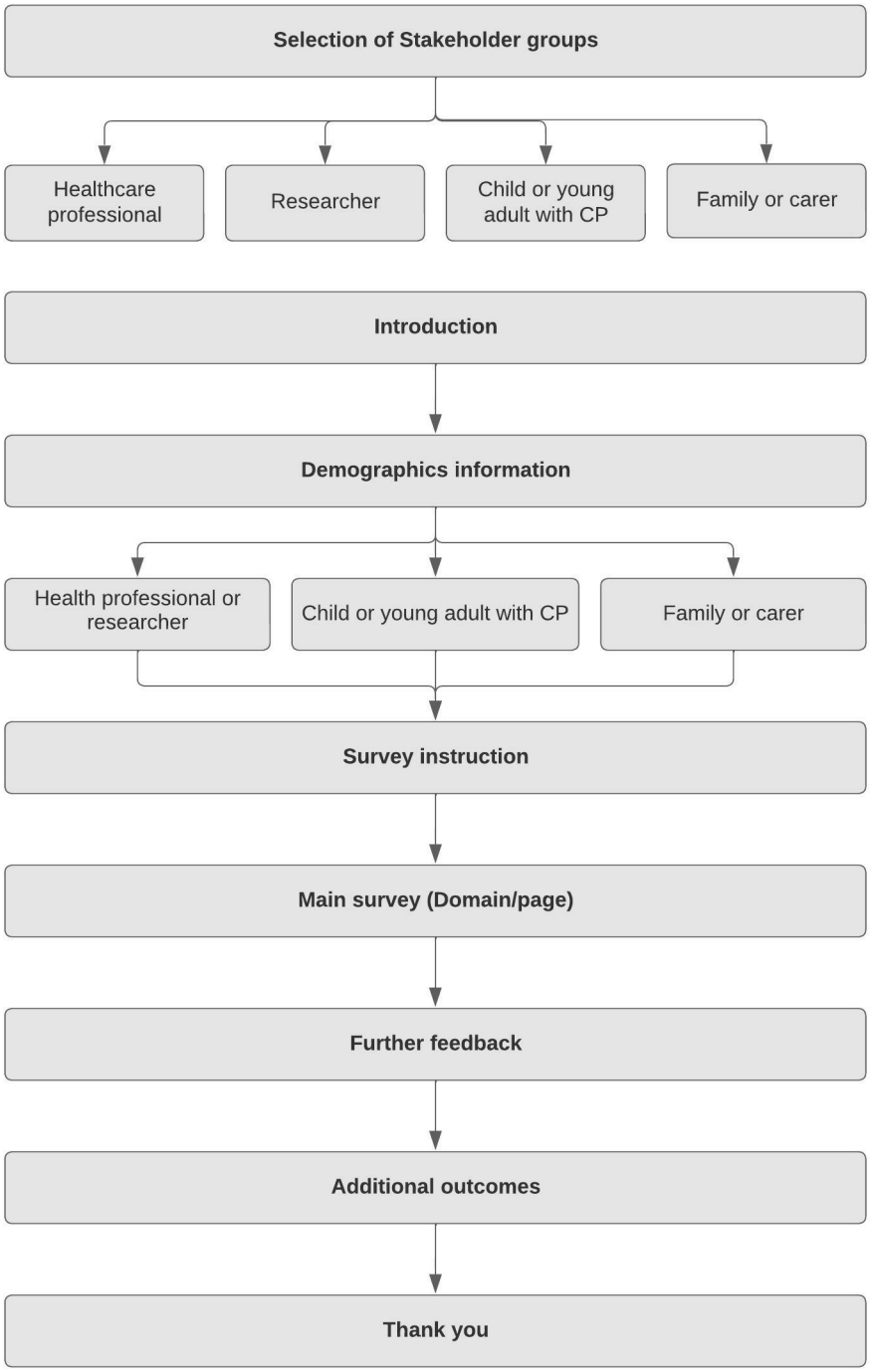


Figure 16 Flowchart illustrating the contents of the e-Delphi survey

Piloting with the steering committee

Emails were sent to members of the project steering committee in which they were asked to pilot and assess the survey for readability, comprehension, clarity and content. A question was added to the survey to ask members to provide any additional outcomes that they considered were missing from the survey. Members were asked to analyse the content and length of the survey and whether they had any

comments about it, including whether they found any sections difficult to understand. For instance, a COS adviser highlighted that the instructions for the survey were not sufficiently clear, and this section was subsequently modified.

9.2.3 Participants and Inclusion Criteria

The COMET guideline (10) advises on the number of stakeholder groups that should be held. They report that the single homogeneous group approach will result in core outcomes that are deemed essential by only one stakeholder group. e.g., patients, who have the clinical condition under study or knowledge of it. A multiple-member, heterogeneous group, within which differing stakeholder opinions are anticipated, has distinct advantages. It is crucial that the final COS must include the outcomes that are deemed essential by all stakeholder groups (9, 10, 215).

For the reasons discussed in Chapter 5, representatives from two key stakeholder panels that might have been particularly interested in lower limb orthopaedic surgery and CP were included. Specific inclusion criteria were defined for each stakeholder panel and are described in Table 44.

Table 44 Participants' inclusion and exclusion criteria

Inclusion Criteria (children, young people and parents)

- Children and young adults:
  o Diagnosed with CP,
  o Ambulant, within GMFCS level I, II, III,
  o Who are aged < 18 years, and adults, who had surgery when they were young,
  o Must have had experience with at least one surgery for any lower limb deformity or have the intention of undergoing that treatment in the future.

- Representatives: o Family member (parents) or carers of a child with above criteria.
Inclusion Criteria (health professionals)

- Clinical researchers:
  o Must have an academic qualification,
  o Be currently employed by a research organisation,
  o have current or 'recent-past' experience in clinical research with studies that focus on questions of clinical efficacy (benefit) of a lower limb orthopaedic surgery or post-surgery-based intervention in children with CP (i.e., authors or a co-author on a relevant peer-reviewed journal publication in the past 10 years).

- Healthcare practitioners:
  o Must have a clinical qualification,
  o be currently employed by a public or private institution that provides a management to children with CP,
  o have specific experience in assessing or managing lower limb musculoskeletal deformity in children with CP who receive, or have received, a surgery, or rehabilitation-based intervention.

General eligibility for participation in the Delphi process included individuals with sufficient ability to read English, and understand and independently complete the questionnaires, except children who may need help from an adult.

9.2.4 Sample Size

There is no agreed statistical method to calculate a sample size for online Delphi surveys and no criteria against which a sample size choice can be judged (497). The sample size depends on the availability of resources and experts in which clinical conditions being studied (9, 10, 215). Murphy et al. (468) highlighted that structured formal consensus methods (e.g., Delphi surveys) often relied on a safety in numbers, as organisers assumed that the greater the number of people involved in the consensus process, the more likely it was that the correct view would be discovered. It was assumed that including as many panel members as possible to represent adequately their corresponding stakeholder group would increase understanding of the important outcomes and enhance the reliability of the group judgement (482, 498-500). In contrast, Akins et al. (501) compared the response characteristics of a homogeneous group of 23 participants against computer-generated samples from 1000 and 2000 participants and found all responses to be stable. The researchers concluded that small, homogeneous groups of Delphi participants (e.g., n=23) could generate reliable responses.

Recent studies in COS development have reported participant numbers between 80 and 150 in round one of modified Delphi surveys. For example, in development of a COS for childhood asthma, n=95 (484), for fibromyalgia, n=96 (502), for acute diarrhoea, n=101 (503) and for cleft palate, n=146 (494). After consideration of previously completed Delphi surveys, a target sample number was set at 100 participants for the groups that would take part in round one. This figure took into account that some participants might drop out between rounds.

9.2.5 Sampling and Recruitment

Healthcare professionals

National and international participants in the healthcare professionals panel were recruited through five methods. First, relevant professionals were contacted and invited to the study via e-mail. Second, national and international associations or societies were asked to disseminate the study information to their membership via e-email (See Appendix 10). Third, a sample of researchers who were identified through a systematic search of the research literature as having previously published one or more articles on CP after lower limb orthopaedic surgery or post-surgical rehabilitation were sent information

about the study. Fourth, the snowball sampling method was used (504, 505), through which existing participants were asked to recruit potential participants with relevant experience who might be interested in participating. Fifth, information with a survey link was posted on social media (e.g., Twitter and Facebook).

Patients and their representatives (parents)

English-speaking children and young adults (aged <18 years) who had been diagnosed with CP previously, and their representatives, who visited the outpatient clinic of a lead NHS site in Oxford for consultations or appointments regarding lower limb surgery, were invited by their paediatric orthopaedic team to participate. The paediatric team informed eligible patients about the study and handed out a patient information sheet in the form of a short animation that explained the purpose of the study (https://www.youtube.com/watch?v=0fvhHgH5V2M). If the child or parents agreed and were willing to participate, a survey link was provided. To ensure that an international perspective of patients and carers was gained, invitations were made through two methods. First, national and international societies were asked to publish newsletter articles and announcements to their members. These societies are listed in Appendix 10. Second, an invitation animation with the survey link was posted on social media (e.g., Twitter and Facebook). For example, the following tweet was shared on Twitter:

@HajarAlmoajil Calls to all children and young adults with cerebral palsy and their carers to complete a 10 min survey, aiming to know which outcomes are important to you after lower limb surgery.
<Survey link>
<Child-friendly animation>

9.2.6 Delphi methodology

The 37 potential outcomes that had been identified formed the basis of the round one survey. The Delphi survey consisted of two rounds (Figure 17). Rounds remained open for eight weeks. Round one was launched on 31 August 2020. Round two was launched on 4 January 2021 with a closing date of 27 February 2021. Regular two-week reminders were sent to those who had partially completed or not completed the survey to maximise response rates.

Round 1 survey

In the first round, participants were asked to consider the importance of each outcome according to whether their lower limb orthopaedic surgery had met its goals, or in the case of participants yet to

undergo surgery, what goals were important to them. As previously stated, participants were asked to score each outcome using the GRADE scale of 1-9, in which 1 represented the least importance and 9 represented the greatest importance (485). If a participant felt that they did not understand a particular outcome, they were able to add feedback. After each outcome and at the end of the round, optional open-text boxes enabled participants to add any comments. Participants were able to propose additional outcome domains that did not appear in the list of outcomes. These additional outcome domains were reviewed and coded, and category labelled by the study team members, with appropriate plain language concepts, to ensure that they represented new items for inclusion in the second round.

Round 2 survey

The COMET guideline (10) states that outcomes that do not reach the pre-defined cut-off in the first round of the Delphi survey may be dropped or retained and represented in the second round. If the initial list of outcomes is large, the inclusion of all outcomes in the second round for rescoring may overburden participants and may increase attrition between the first and second rounds.

Hence, in the second round of this Delphi survey, outcomes that had reached the pre-specified criteria (explained in detail in section Survey Development) of 'consensus in' or 'consensus out' in the first round were not taken forward to the second round. Outcomes with the status of 'no consensus', and the additional outcomes suggested by participants, were included for rating by participants in the second round. The percentage scores for each outcome in round one were provided to the stakeholder groups. Participants were asked to re-score the importance of each outcome. Participants were asked to consider which outcomes were of core importance and the need for a COS that comprised five to ten outcomes was emphasised.

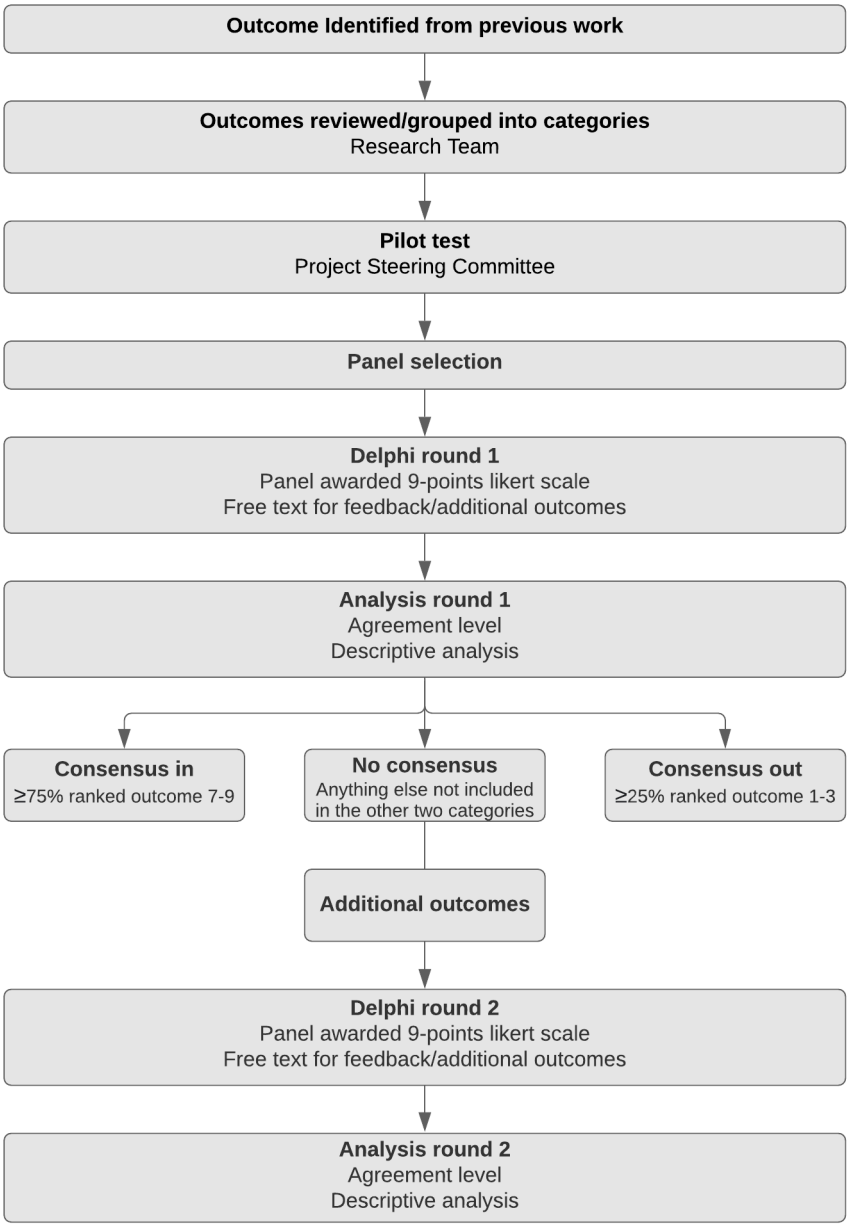


Figure 17 Flow chart illustrating the selection of outcomes throughout the Delphi process

9.2.7 Minimisation of attrition

Attrition bias may occur if participants do not take part in subsequent rounds of survey (10). There is little evidence regarding the impact of attrition bias in COS studies and there is no guidance on what constitutes an adequate response rate. However, Goodman (506) emphasised the importance of minimising attrition in Delphi studies, because people with minority opinions were more likely to drop out of the process and this can lead to an overestimation of the degree of consensus in the final results.

Attrition rates vary between studies and seem to depend to some degree on the method of recruitment; it is assumed that attrition rates are higher when fewer recruitment approaches are used. Thus, in this study, various issues were considered to minimise this effect. First, the information provided with the survey emphasised the importance of completing both rounds; this was emphasised again when the second-round survey was distributed. Second, so that people felt that their voices were being heard, in the invitation to complete round two we acknowledged the differences in how people had voted in round one. Third, various recruitment approaches were used, as presented in section 9.2.5.

9.2.8 Ethical management and data protection

Consent

Consent was implied when the participant registered their name and email address to take part in the Delphi survey via the website. This is in line with Health Research Authority (HRA) guidance: "Studies with little or no intervention and less than minimal risk is likely to need a much shorter information sheet and you will not need to complete all sections (for example the explanation of a questionnaire study may be summarised on the front of the questionnaire itself and completion of the questionnaire regarded as consent)" (HRA Guidance, page 34). In addition, at the start of the survey, participants were asked to tick a box to indicate consent.

Anonymity

No clinical information on participants was collected or stored. When participants registered to take part in the Delphi survey, they were asked to register an email address so that reminders to complete the survey could be sent appropriately. Survey responses were anonymised by allocation of a unique participant number to each participant, and they could not be traced not traceable back to the respondent except by the immediate research team.

Data protection

Data were handled in compliance with the Data Protection Act (1998). Records linking individual pieces of data to the participant number were kept in a password-protected document on a secure server at the University of Oxford that could be accessed by the immediate research team only. In line with the university's policy, data will be archived at the University of Oxford for at least five years. After this period, the data will be destroyed.

9.2.9 Data Analysis

Descriptive statistics were used to describe the stakeholder groups and the results for each outcome. Quantitative analysis of the Delphi survey included calculations of each panel's response rate as percentages and the percentage of responses in each of the three importance categories (1-3: 'not important', 4-6: 'important but not critical' and 7-9: important and critical') based on the nine-point Likert scale for each outcome. This scale can be assumed to be an interval scale; therefore, descriptive statistics such as the median and interquartile range (IQR) were calculated for each outcome at the end of each round.

The median was used as a measure to express the average opinion of the panel. If the median was high, then the panel was in strong agreement about the importance of the outcome; if the median was low, then there was disagreement within the panel. Therefore, the median of each outcome was reported in order to assess any tendency towards agreement (i.e., opinion stability) for each stakeholder group across the Delphi rounds. The IQR for each outcome, which was calculated as the difference between the third and the first quartiles, was reported to assess the extent of agreement between the participants. The data were reported in table form and graphically in box-plot graphs for each stakeholder group separately.

There is currently no guidance on exactly how the views of different stakeholder groups should be handled. It is recognised that the way in which the views of different groups of participants are combined in a single heterogeneous panel can greatly influence the results of a Delphi process (215). The criteria for consensus are based on the group overall and, where there is variation in the numbers of participants in different groups, the views of small groups are likely to be overwhelmed, i.e., consensus is likely be weighted towards the views of a particular stakeholder group. Separation of these groups into multiple homogeneous panels offers distinct advantages in that it preserves the opinions of minority groups. In accordance with other studies, it was decided to analyse the results of stakeholder groups separately throughout the Delphi process, and then to identify important outcomes based on the consensus criteria of each group and across stakeholder groups that had prioritised them (507).

To examine whether there was evidence that bias may have been introduced through participant attrition in the second round, the average scores after the first round were compared for those who completed the next round and those who had dropped out after the first round.

9.3 Results

9.3.1 Expert panel (stakeholders)

Given the nature of the study recruitment procedure, it is not possible to know how many people received invitations to participate during the window of recruitment. There were 111 respondents to the first-round survey. Stakeholders were placed into two groups. Group one consisted of health professionals (n=93) and group two comprised of individuals with CP and their representatives (n=18). The second round was completed by 86 participants, 68 of whom were healthcare professionals, and 18 of whom were individuals with CP and their representatives. Therefore, 66 respondents (59.4%) who had completed round one also completed round two.

Group one members included orthopaedic surgeons, paediatricians, physiotherapists, and researchers. Others represented a variety of healthcare backgrounds: one was a paediatric physiatrist, one was a human movement scientist, one was an orthotist and one was a nurse. In group two, of those with CP who took part in the first round, most were older than 18 years of age, while most of those with CP who took part in the second round were aged between 11 and 15 years old. Their representatives (parents) were mainly mothers of a child with CP. The representation was from eight world regions and 13 countries (Figure 18). The majority of respondents were from the UK and Australia. Tables 45 and 46 outline the numbers and characteristics of the participants within each group in survey rounds one and two.

Table 45 Characteristics of the healthcare professionals in the e-Delphi survey rounds one and two [n (%)]

	Round 1	Round 2
	n= 93	n= 68
Current professionals		
Physiotherapists	44 (47.3)	24 (35.2)
Orthopaedic surgeons	34 (36.5)	32 (47)
Researchers	9 (9.6)	10 (14.7)
Paediatricians	2 (2.1)	1 (1.4)
Nurses	1 (1)	-
Paediatric physiatrists	1 (1)	-
Human movement science	1 (1)	-
Orthotist	1 (1)	1 (1.4)
Years of experience		
> 10 years	58 (62.3)	46 (67.6)
5-10 years	20 (21.5)	16 (23.5)
< 5 years	15 (16.1)	6 (8.8)
Country of origin		
United Kingdom	33 (35.4)	20 (29.4)
Australia	22 (23.6)	14 (20.5)
Asia	15 (16.1)	11 (16.1)
Europe	12 (12.9)	13 (19.1)
United States	6 (6.4)	6 (8.8)
South America	3 (3.2)	2 (2.9)
Canada	1 (1)	1 (1.4)
Africa	1 (1)	1 (1.4)
Gender		
Female	61 (65.5)	38 (55.8)
Male	31 (33.3)	29 (42.6)
Prefer not to say	1 (1)	1 (1.4)

Table 46 Characteristics of the individuals with CP and representatives in the e-Delphi survey rounds one and two [n (%)]

	Round 1		Round 2	
	n= 18		n= 18	
	Patients	**Representatives**	**Patients**	**Representatives**
Country of origin				
United Kingdom	2 (11.1)	5 (27.7)	3 (16.6)	6 (33.3)
Australia	3 (16.6)	1 (5.5)	2 (11.1)	3 (16.6)
Asia	-	-	-	-
Europe	-	-	-	-
United States	2 (11.1)	1 (5.5)	2 (11.1)	1 (5.5)
South America	2 (11.1)	-	-	-
Canada	-	-	-	-
Africa	-	2 (11.1)	-	1 (5.5)
Gender				
Female	4 (22.2)	7 (38.8)	2 (11.1)	8 (44.4)
Male	5 (27.7)	2 (11.1)	4 (22.2)	3 (16.6)
Prefer not to say			1 (5.5)	
Child's age				
6-10 years	-	4 (22.2)	-	2 (11.1)
11-15 years	1 (5.5)	1 (5.5)	3 (16.6)	6 (33.3)
16-18 years	1 (5.5)	1 (5.5)	1 (5.5)	-
> 18 years	7 (38.8)	3 (16.6)	3 (16.6)	3 (16.6)
Time since surgery				
Not had surgery yet	1 (5.5)	2 (11.1)	1 (5.5)	2 (11.1)
< 6 months	-	3 (16.6)	-	1 (5.5)
6-12 months	1 (5.5)	1 (5.5)	-	-
1-3 years	-	-	3 (16.6)	5 (27.7)
3-5 years	2 (11.1)	1 (5.5)	1 (5.5)	1 (5.5)
> 5 years	5 (27.7)	2 (11.1)	2 (11.1)	2 (11.1)
Child's current walking ability indoors				
Most of the time, they walk on their own	4 (22.2)	3 (16.6)	5 (27.7)	6 (33.3)
Most of the time, they walk using a crutch or walker	-	1 (5.5)	1 (5.5)	2 (11.1)
Most of the time, they walk with my support	3 (16.6)	3 (16.6)	1 (5.5)	1 (5.5)
Most of the time, they use a wheelchair	2 (11.1)	2 (11.1)	-	2 (11.1)
Child's current walking ability outdoor				
Most of the time, they walk on their own	3 (16.6)	1 (5.5)	2 (11.1)	3 (16.6)
Most of the time, they walk using a crutch or walker	-	3 (16.6)	2 (11.1)	2 (11.1)
Most of the time, they walk with my support	2 (11.1)	1 (5.5)	2 (11.1)	3 (16.6)
Most of the time, they use a wheelchair	4 (22.2)	4 (22.2)	1 (5.5)	3 (16.6)

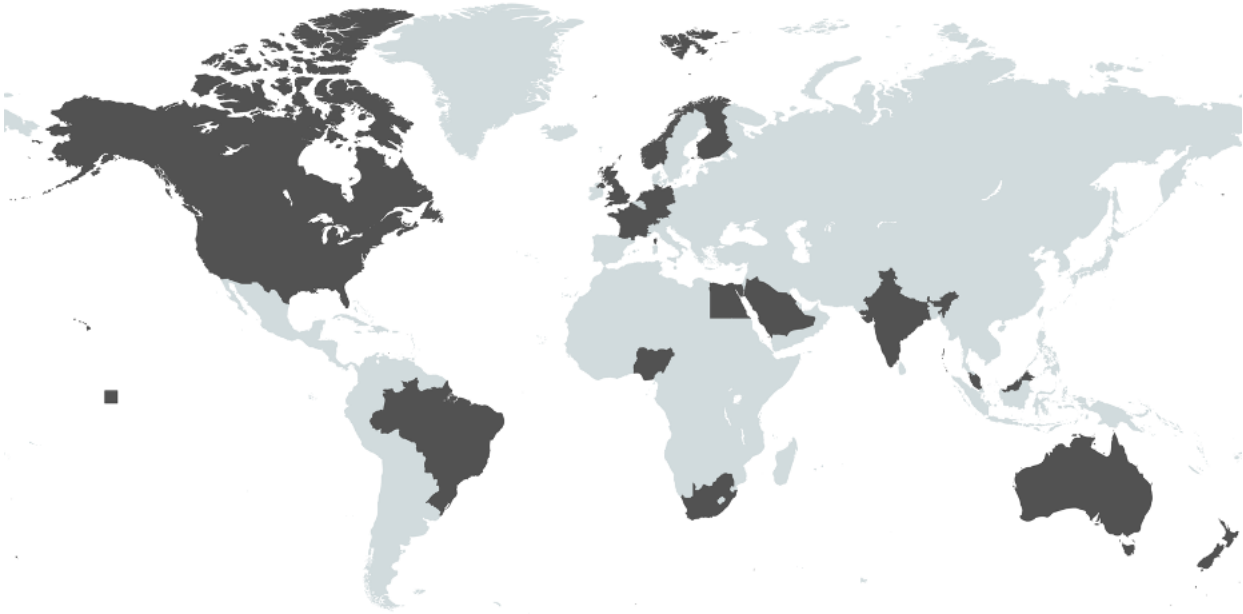


Figure 18 Geographical distribution of Delphi participations

9.3.2 Round One

Table 47 shows the numbers and percentages of first round participants who scored each outcome as 1-3, 4-6 and 7-9 on the nine-point scale. Analysis of the survey by stakeholder group showed that there was heterogeneity in the number of outcomes that reached consensus; 18 outcomes amongst health professionals and 12 outcomes amongst individuals with CP and representatives. Health professionals explained in the feedback section that all the outcomes were critical for their patients at some point after treatment and for children at different functional levels and ages; this information shed light on why health professionals rated few outcomes at below seven points ('not important' or 'important but not critical') on the scale. Two outcomes reached consensus of limited importance (i.e., consensus out) according to the group of individuals with CP and representatives. These outcomes were related to lower limb symmetry and aesthetic goals.

Consensus across stakeholders' panels

Consensus across all panels was used to define the second round of the Delphi survey. A total of 11 outcomes (30%) met the 75% threshold and were classified as 'consensus in' from both stakeholder groups; none were classified as 'consensus out'; and 26 (70%) were classified as 'no consensus'. Among the 11 outcomes that were classified 'consensus in' across both stakeholder groups, three common domains were identified: avoiding falls; life impact (walking and independence); and delivery of care that may have an impact on the surgical decision. Table 47 and Figures 19 and 20 outline the median score and IQR for each outcome based on the responses to round one of the survey.

Suggested outcome domains

An additional six outcomes were suggested by respondents in round one. Of these, two outcomes were considered to have been included in the existing questions. Four were regarded as unique outcomes and were re-worded in plain language and included in round two for scoring. These were: to be able to join in with more activities at school (e.g., physical education (PE)); to be able to stand with a better balance; to be able to walk with the foot flat on the ground; and ability to adhere to home exercises. Participants were informed of the new outcomes and asked to rank these alongside the other outcomes in round two.

Table 47 Descriptive analysis of outcomes included in the Delphi study round one

	Health professionals (n= 93)					Individuals with CP/ Representatives (n= 18)				
	N (%)			M	IQR	N (%)			M	IQR
	1-3	4-6	7-9			1-3	4-6	7-9		
To decrease the child's pain and muscle soreness										
To feel less pain	4 (4)	9 (10)	80 (86)	8	(7 - 9)	2 (12)	3 (18)	12 (71)	8	(6 - 9)
To feel less muscle soreness or tiredness after activities	6 (6)	25 (27)	62 (67)	7	(6 - 8)	3 (18)	5 (29)	9 (53)	8	(5 - 9)
To improve the way the child looks										
To be able to stand taller or more upright	7 (8)	30 (33)	55 (60)	7	(6 - 8)	2 (12)	4 (24)	11 (65)	7	(6 - 9)
To have straighter and better shaped legs	10 (11)	42 (46)	40 (43)	6	(5- 7)	3 (17)	4 (22)	11 (61)	7	(5.25 - 8.75)
To have legs that look the same as each other	16 (17)	43 (46)	34 (37)	6	(4- 7)	5 (29)	1 (6)	11 (65)	7	(3 - 9)
To increase how much the child can do										
To reduce or prevent falls	1 (1)	15 (16)	77 (83)	8	(7 - 9)	1 (6)	-	17 (94)	9	(8 - 9)
To be able to climb stairs	5 (5)	34 (37)	53 (58)	7	(6 - 8)	2 (11)	5 (28)	11 (61)	8	(6 - 8.75)
To be able to do more activities at home	1 (1)	8 (9)	84 (90)	8	(7 - 9)	1 (6)	4 (22)	13 (72)	8	(6.5 - 9)
To improve participation in sports (e.g., football, swimming)	3 (3)	26 (28)	64 (69)	7	(6 - 8)	4 (22)	5 (28)	9 (50)	6.5	(4.25 - 8.75)
To improve participation in hobbies (e.g., dancing, horse-riding)	2 (2)	18 (19)	73 (78)	8	(7 - 8)	2 (12)	5 (29)	10 (59)	8	(4 - 9)
To increase the child's independence										
To be able to go out of the house independently (without help from an adult)	3 (3)	8 (9)	82 (88)	8	(7 - 9)	2 (11)	1 (6)	15 (83)	8	(7 - 9)
To be able to do things independently at home	1 (1)	8 (9)	84 (90)	8	(7 - 9)	2 (11)	1 (6)	15 (83)	9	(8 - 9)
To be able to do activities of daily living independently (e.g., bathing, dressing and self-care)	1 (1)	8 (9)	84 (90)	8	(7 - 9)	2 (11)	1 (6)	15 (83)	9	(8.25 - 9)
To be able to walk without assistance when going out	2 (2)	22 (24)	67 (74)	7	(6 - 9)	4 (22)	4 (22)	10 (56)	9	(5.25 - 9)
To be able to walk without assistance at home	1 (1)	15 (16)	75 (82)	8	(7 - 9)	3 (18)	1 (6)	13 (76)	9	(7 - 9)
To improve the way the child walks after surgery										
To be able to walk with straight legs	3 (3)	41 (45)	48 (52)	7	(5.75- 8)	3 (17)	4 (22)	11 (61)	8	(6 - 9)
To be able to walk without getting tired	2 (2)	14 (15)	77 (83)	8	(7 - 8)	1(6)	3 (17)	14 (78)	8.5	(7 - 9)
To be able to walk faster	5 (5)	35 (38)	52 (57)	7	(6 - 8)	1 (6)	9 (50)	8 (44)	6	(6 - 7.75)
To be able to walk further	2 (2)	18 (19)	73 (78)	7	(7 - 8)	2 (11)	2 (11)	14 (78)	7	(7 - 8.75)

To be able to walk without using a crutch or walker	6 (7)	41 (45)	44 (48)	6	(5 - 7)	4 (22)	1 (6)	13 (72)	9	(6.25 - 9)
To be able to walk better even if continue using a crutch or walker	1 (1)	9 (10)	82 (89)	8	(7 - 8)	4 (22)	2 (11)	12 (67)	8	(5.25 - 9)
To stop wearing a brace /splint/ orthosis	13 (14)	41 (44)	39 (42)	6	(5 - 7)	4 (22)	4 (22)	10 (56)	7.5	(4 - 9)
To improve how the child feel when interacts with other people										
The child improves friendships and relationships with other people	11 (12)	32 (34)	50 (54)	7	(5 - 8)	4 (22)	9 (50)	5 (28)	5	(4.25 - 7.5)
To feel more confident	5 (5)	20 (22)	67 (73)	7	(6 - 9)	3 (17)	1 (6)	14 (78)	7.5	(7 - 9)
To feel happier	6 (6)	22 (24)	65 (70)	7	(6 - 9)	3 (17)	3 (17)	12 (67)	8	(6 - 9)
To feel better about how other people treat the child	11 (12)	27 (30)	52 (58)	7	(5 - 8)	6 (33)	3 (17)	9 (50)	6.5	(3 - 7.75)
Concerns about having surgery										
Feeling pain after the surgery	6 (6)	27 (29)	60 (65)	7	(6 - 8)	2 (11)	3 (17)	13 (72)	8	(6.25 - 8.75)
Developing wound infection	4 (4)	39 (42)	50 (54)	7	(5 - 8)	3(17)	4 (22)	11 (61)	8	(4.25 - 8.75)
Losing independence	3(3)	15 (16)	75 (81)	8	(7 - 9)	4 (22)	6 (33)	8 (44)	6	(4 - 8)
Needing to have another surgery	2 (2)	35 (38)	55 (60)	7	(6 - 8)	1 (6)	6 (33)	11 (61)	7.5	(6 - 8)
Getting worse after surgery	3 (3)	11 (12)	78 (85)	8	(7 - 9)	1 (6)	4 (22)	13 (72)	7.5	(6.25 - 9)
Taking a long time to walk again	3 (3)	25 (27)	65 (70)	7	(6 - 8)	2 (11)	4 (22)	12 (67)	8	(5 - 9)
How important are the following factors in your decision to recommend the surgery?										
Good access to physiotherapy services locally	1 (1)	10 (11)	82 (88)	8	(7 - 9)	1 (6)	1 (6)	16 (89)	9	(8 - 9)
Good communication with other healthcare professionals	2 (2)	10 (11)	81 (87)	8	(7 - 9)	-	2 (11)	16 (89)	9	(8 - 9)
Being well informed about the operation and potential risks	-	6 (6)	87 (94)	8	(8 - 9)	-	-	18 (100)	9	(8.25 - 9)
Getting help from the right health professionals when needed	-	6 (6)	87 (94)	8	(7 - 9)	-	2 (11)	16 (89)	9	(8 - 9)
Getting support from family/friends	-	8 (9)	85 (91)	8	(7 - 9)	-	7 (39)	11 (61)	7.5	(6 - 9)

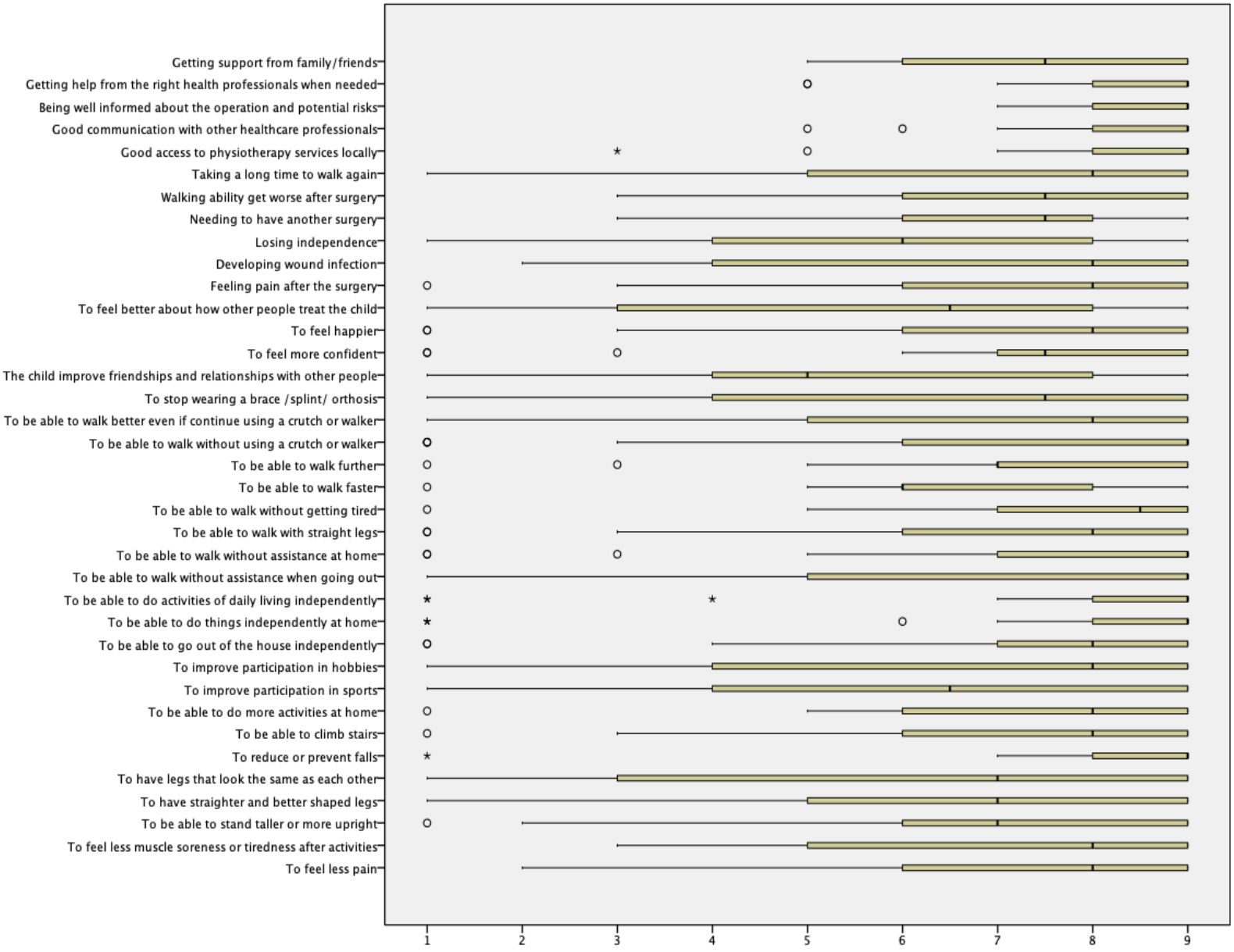


Figure 19 Distribution of median and IQR for each outcome (health professionals)- round one

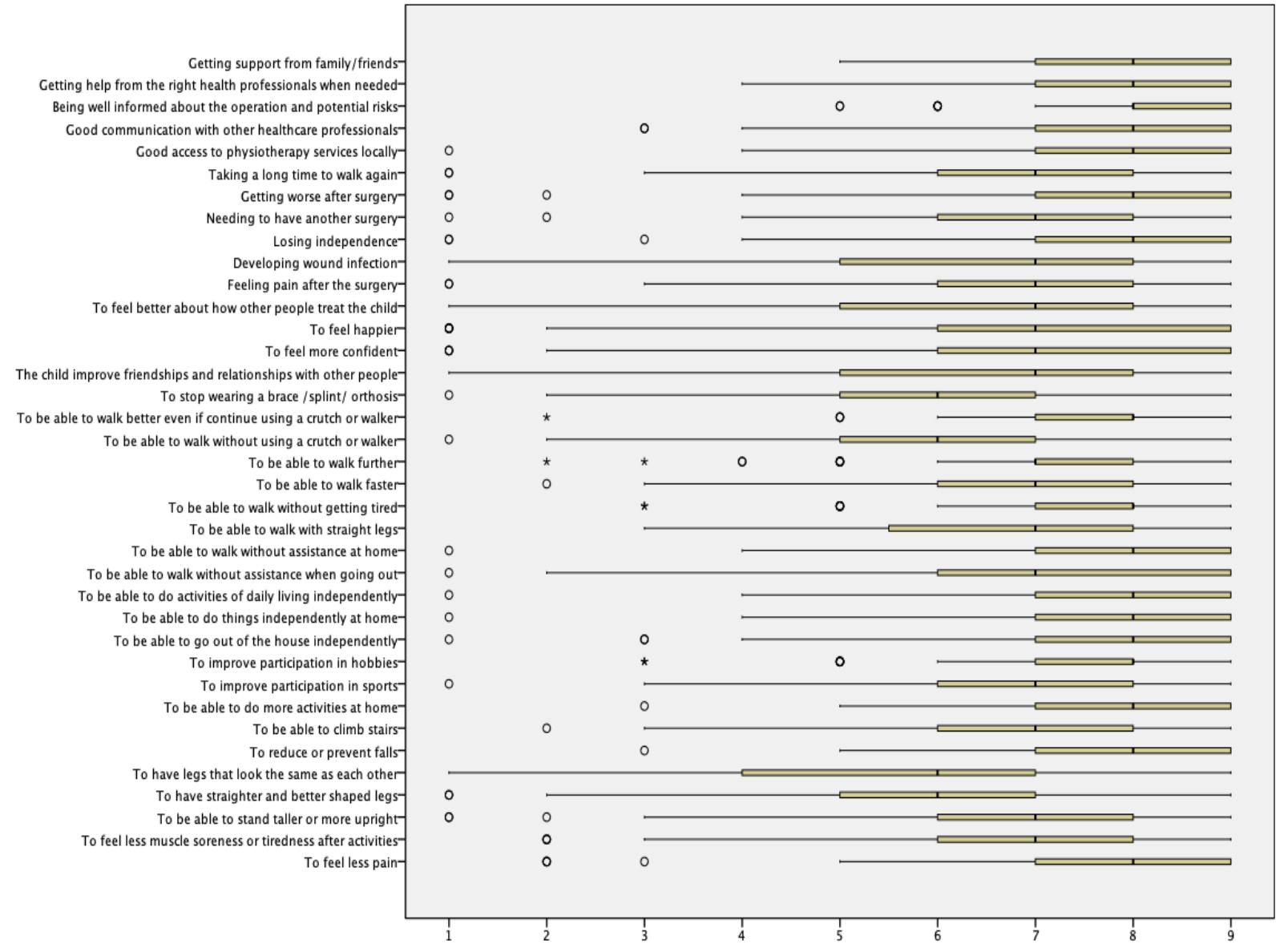


Figure 20 Distribution of median and IQR for each outcome (individuals with CP/ representatives)- round one

9.3.3 Round two

In the second round, participants were asked to rate 30 outcomes. As in the round one survey, there was heterogeneity in the number of outcomes that reached consensus (Table 48). Among health professionals, 12 outcomes reached consensus whereas, among individuals with CP and representatives, nine did. Similar to round one, two outcomes reached 'consensus out' among individuals with CP and representatives, which were: improve participation in PE activities at school and one of the adverse effects of surgery; symptoms worsen after the operation.

Consensus across stakeholders' panels

Across all stakeholder groups, five outcomes (16.6%) met the 75% threshold and were classified as 'consensus in'; none were classified as 'consensus out'; and 25 (83.4%) were classified as 'no consensus'. Among the five outcomes that reached consensus, three core areas were identified: avoiding pain; mobility and life impact (daily life activities); independence; and delivery of care that might have impact on the decision to undergo surgery. Table 48 and Figures 21 and 22 outline the median score and IQR for each outcome based on the responses to the round two survey. No further outcomes were suggested in this round.

Table 48 Descriptive analysis of outcomes included in the Delphi survey round two

	Health professionals (n= 68)					Individuals with CP/ Representatives (n= 18)				
	N (%)			M	IQR	N (%)			M	IQR
	1-3	4-6	7-9			1-3	4-6	7-9		
To decrease the child's pain and muscle soreness										
To feel less pain	1 (1)	9 (13)	58 (85)	8	(7 - 9)	2 (12)	1 (6)	14 (82)	8	(7 - 9)
To feel less muscle soreness or tiredness after activities	1 (1)	23 (34)	44 (65)	7	(6 - 8)	-	3 (17)	15 (83)	8	(7 - 9)
To improve the way the child looks										
To be able to stand taller or more upright	3 (4)	21 (31)	44 (65)	7	(6 - 8)	4 (24)	2 (12)	11 (65)	7	(5 - 9)
To have straighter and better shaped legs	5 (7)	41 (60)	22 (32)	6	(5 - 7)	2 (11)	3 (17)	13 (72)	8	(5.5 - 9)
To have legs that look the same as each other	13 (19)	39 (57)	16 (24)	5	(4 - 6)	4 (24)	3 (18)	10 (59)	7	(4 - 8)
To increase how much the child can do										
To be able to climb stairs	1 (1)	31 (46)	36 (53)	7	(6 - 8)	2 (11)	5 (28)	11 (61)	7.5	(5 - 8)
To be able to do more activities at home	-	8 (12)	60 (88)	8	(7 - 9)	1 (6)	2 (11)	15 (83)	8	(7 - 9)
To improve participation in sports (e.g., football, swimming)	1 (1)	26 (38)	41 (60)	7	(6 - 8)	3 (17)	4 (22)	11 (61)	7	(5.2.5 - 9)
To improve participation in hobbies (e.g., dancing, horse-riding)	1 (1)	19 (28)	48 (71)	7	(6 - 8)	3 (17)	3 (17)	12 (67)	8.5	(6 - 9)
To be able to join more activities at school (e.g., PE)*	-	12 (18)	56 (82)	8	(7 - 8)	5 (28)	1 (6)	12 (67)	7	(3.5 - 9)
To be able to stand with a better balance*	-	9 (13)	59 (87)	8	(7 - 9)	2 (11)	-	16 (89)	9	(8 - 9)

To increase the child's independence										
To be able to walk without assistance when going out	2 (3)	15 (22)	51 (75)	8	(6.75 -8)	2 (11)	0	16 (89)	9	(7.25 - 9)
To improve the way the child walks after surgery										
To be able to walk with straight legs	1 (1)	24 (35)	43 (63)	7	(6 - 8)	2 (11)	1 (6)	15 (83)	8	(8 - 9)
To be able to walk faster	3 (4)	24 (35)	41 (60)	7	(6 - 8)	3 (17)	4 (22)	11 (61)	7	(6 - 8)
To be able to walk without using a crutch or walker	3 (4)	30 (45)	34 (51)	7	(5 - 8)	3 (17)	3 (17)	12 (67)	8	(6 - 9)
To be able to walk better even if continue using a crutch or walker	-	8 (12)	60 (88)	8	(7 - 9)	3 (17)	4 (22)	11 (61)	8	(5.25 - 8.75)
To stop wearing a brace /splint/ orthosis	14 (21)	28 (42)	24 (36)	6	(5 - 7)	3 (17)	1 (6)	14 (78)	8.5	(7 - 9)
To be able to walk with the foot flat on the ground*	2 (3)	24 (35)	42 (62)	7	(6 - 8)	1 (6)	1 (6)	16 (89)	9	(8 - 9)
To improve how the child feel when interacts with other people										
The child improves friendships and relationships with other people	5 (7)	27 (40)	36 (53)	7	(5 - 8)	2 (11)	9 (50)	7 (39)	6	(5 - 8)
To feel more confident	3 (4)	9 (13)	56 (82)	7.5	(7 - 8)	2 (11)	4 (22)	12 (67)	9	(6 - 9)
To feel happier	2 (3)	12 (18)	54 (79)	8	(7 -8.75)	1 (6)	4 (22)	13 (72)	9	(6.25 - 9)
To feel better about how other people treat the child	6 (9)	27 (40)	35 (51)	7	(5 - 8)	4 (22)	5 (28)	9 (50)	6.5	(5 - 9)
Concerns about having surgery										
Feeling pain after the surgery	2 (3)	20 (29)	46 (68)	7	(6 - 8)	1 (6)	5 (28)	12 (67)	7	(6 - 9)
Developing wound infection	3 (4)	26 (38)	39 (57)	7	(6 - 8)	4 (22)	9 (50)	5 (28)	6	(4.25 - 6.75)
Losing independence	2 (3)	5 (7)	61 (90)	8	(7 - 9)	3 (17)	4 (22)	11 (61)	7.5	(6 - 8.75)
Needing to have another surgery	5 (7)	21 (31)	42 (62)	7	(6 - 8)	2 (11)	8 (44)	8 (44)	6	(5 - 7.75)
Getting worse after surgery	2 (3)	5 (7)	61 (90)	8	(7 -8.75)	5 (28)	5 (28)	8 (44)	6	(3.25 - 9)
Taking a long time to walk again	2 (3)	21 (31)	44 (66)	7	(6 - 8)	3 (17)	6 (33)	9 (50)	6.5	(5 - 9)
How important are the following factors in your decision to recommend the surgery?										
Getting support from family/friends	1 (1)	12 (18)	55 (81)	8	(7 - 9)	2 (11)	3 (17)	13 (72)	7	(6.25 - 9)
Ability to adhere to home exercises*	2 (3)	10 (15)	56 (82)	8	(7 - 9)	1 (6)	2 (11)	15 (83)	9	(7 - 9)

*New suggested outcomes from previous round M: median IQR: interquartile range

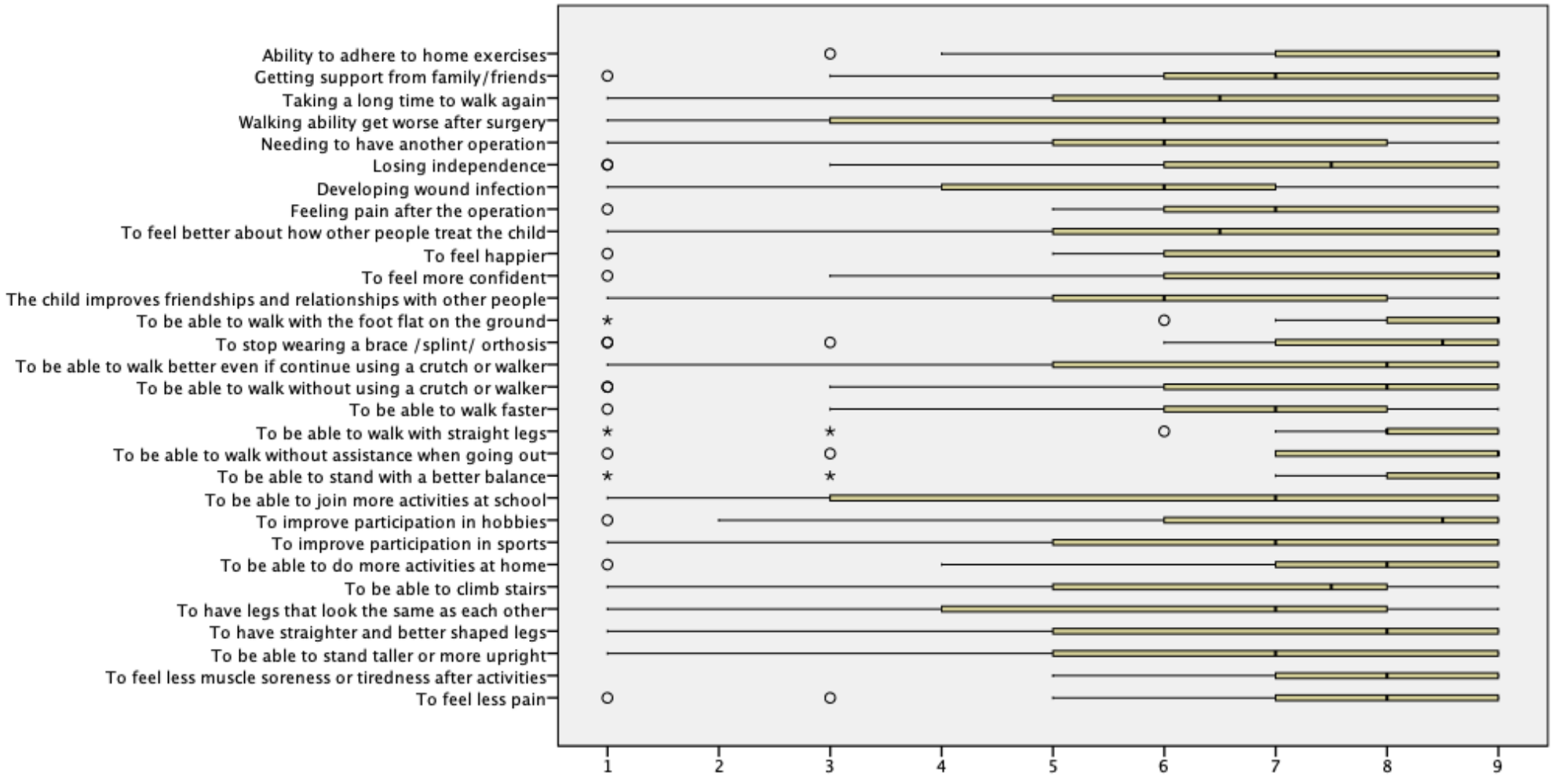


Figure 21 Distribution of median and IQR for each outcome (health professionals)- round two

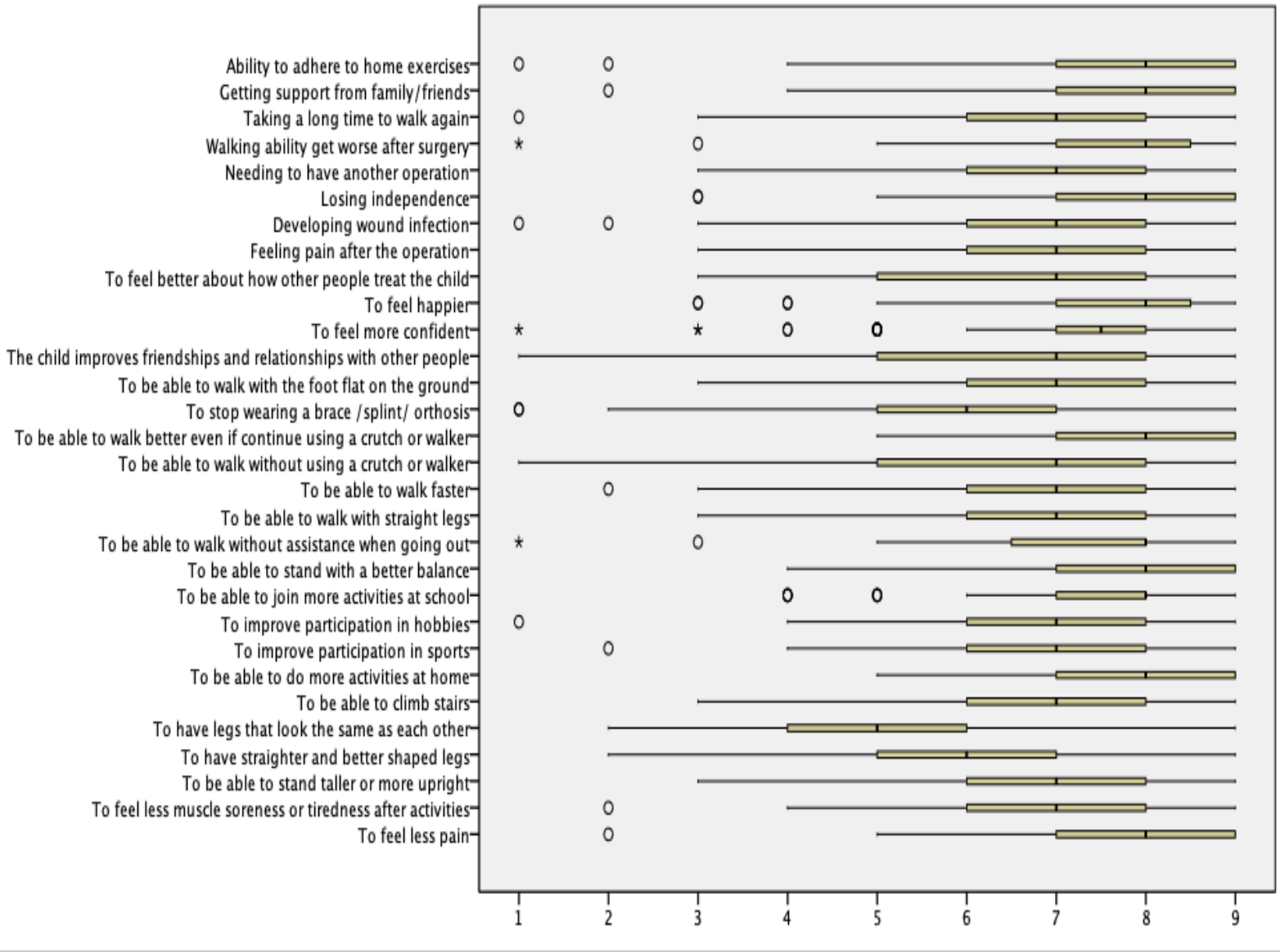


Figure 22 Distribution of median and IQR for each outcome (individuals with CP/ representatives)- round two

9.3.4 Rounds one and two

On review of both rounds, the results were outlined according to themes. These were: outcomes deemed important and critical, 'consensus in' across stakeholder groups (n= 16); outcomes deemed important for each stakeholder group, health professionals (n= 8) and individuals with CP and representatives (n= 4); outcomes ranked as not important by individuals with CP and representatives (n= 4); and finally, outcomes that reached 'no consensus' level (n= 11). Table 49 illustrates the level of consensus of outcome domains across the first and second rounds of the Delphi survey per stakeholder group. Based on the percentage scores, outcomes were categorised into each level of importance and organised into consensus categories **A:** outcomes that reached *consensus in* by both or either group, **B:** outcomes that reached *consensus out* by both or either group, or **C:** outcomes that reached *no consensus* by both groups.

Outcomes that reached 'consensus in' across both stakeholder groups (n= 16)

There was substantial coherence between stakeholder groups, with 16 outcomes achieving the 75% threshold of importance and labelled as 'consensus in' across stakeholder groups. These outcomes were categorised into the broad domains: physiological, motor function, life impact (e.g., mobility, social life and independence) and quality of life outcomes, and delivery of health services that may impact the decision to undertake surgery.

Within the physiological outcome category, avoidance of pain was rated important and critical and was, therefore, to be included in the final COS. Health professionals maintained their rating of its importance between rounds, while the rating increased between rounds among individuals with CP and representatives, from 71% in round one to 82% in round two. Two outcomes reached a consensus in the motor function health domain; these were avoiding the incidence of falls and maintenance of static balance. The former reached a high level of consensus across both groups: health professionals (83%) and individuals with CP and representatives (94%). The latter was suggested by the health professionals panel in the first round and its importance was agreed by both groups in the second round.

The outcome areas that were related to life impact also reached beyond for inclusion for the COS. They were walking-related outcomes such as increased walking endurance and lengthened distance. Those outcomes that reached a high level of consensus in the life impact domain were related to independence and were: being independent in daily life activities (e.g., self-care) and both indoor and outdoor

activities, and being able to walk independently without assistance from an adult within the home and the community. All the life impact outcomes reached consensus rates from 75% to 90%. In the quality of life domain, only the confidence outcome met the threshold of 'important' according to all stakeholder groups. However, the rating of this outcome by individuals with CP and representatives dropped from 'consensus in' (78%) in the first round to 'no consensus' in the second round.

Finally, factors related to the ability of health services to deliver the required care, which could have an impact on decision to undertake the surgery, reached a high level of consensus amongst all stakeholder groups. Outcomes that reached consensus in this domain were: access to physiotherapy services; communication and support from healthcare professionals; availability of information resources; and the ability of the child to adhere to home exercises. The rates of consensus for these outcomes ranged from 81% to 100%.

Outcomes that reached 'consensus in' by health professionals (n= 8)

The healthcare professionals' panel rated seven outcomes as important. It has been found previously that health professionals do not consider that surgery will change a child's level of gross motor function classification system. In line with this, they stated several times in the feedback that change to gross motor function 'depends on the pre-operative ability'. Thus, they rated improvement of the child's walking ability even if the child used as assistive device as an important goal of surgery in rounds one and two (89% and 88%, respectively).

The health professionals rated the social life outcomes of participation in leisure and hobbies such as dancing or horse-riding as important in the first round (78%), but this rating was reduced to 'no consensus' after round two (71%). This change was due to participation by fewer health professionals in the second round, which led to a fall in the rating below the 75% threshold for inclusion. In contrast, participation in school activities (e.g., PE) with peers, which was added as an outcome after round one, was rated as important and critical for postoperative assessment in round two (82%). Satisfaction with the surgical outcomes that represented quality of life was also rated as of critical importance (79%).

Two possible adverse events after surgery, loss of independence and worsened walking ability, were rated 7-9 by 90% of health professionals in the second round. Furthermore, health professionals also appeared to have reflected and increased their agreement between the rounds regarding the

importance of support from family or friends during the post-surgery recovery period. This rating increased from 81% in the first round to 91% in the second round.

Outcomes that reached 'consensus in' by individuals with CP and representatives (n= 4)

Four outcomes reached the pre-defined criteria of 'consensus in' according to the young people and representatives. One of the physiological outcomes, reduction of fatigue, reached an 83% rating in the second round as assessed by this group. Other outcomes that were agreed to be important were related to the life impact core area, such as the ability to walk better without using braces, splints or orthoses. Although health professionals suggested in the feedback to the first round that 'walking with foot flat on the ground' should be added as a new outcome, it was rated as important and critical by only individuals with CP and their representatives (89%).

Outcomes that reached 'consensus out' (n= 4)

'Consensus out' ratings were markedly different between the two groups. The health professionals did not rate any single outcome as of limited importance. In contrast, for the young people and representatives group, four outcomes met the 25% threshold of limited importance: lower limb symmetry (29%); participation in school activities (e.g., PE) (28%), which was rated as 'consensus in' by the health professionals' group; aesthetic, which was rated 1-3 (not important) by 33% of the group in the first round but less than 25% in the second round; and worsened walking ability after the operation was rated as not important (28%), whereas 90% of health professionals rated this outcome as of high importance.

Outcomes reached 'no consensus' across stakeholder groups (n= 11)

Among a total of 41 outcome domains, 11 reached 'no consensus' after results from both groups had been combined. Two outcomes were related to lower limb symmetry and alignment. Three were from the life impact domains: the ability to climb stairs; the ability to walk without the use of assistive devices; and the ability to walk faster than the child could before the surgery. One outcome was linked with social life: the ability to participate in sport (e.g., football, swimming). Another was improved socialisation with friends and peers after surgery, which represented the quality-of-life domain. Finally, no consensus was reached on four outcomes related to adverse events that could occur after surgery: postoperative pain syndrome, wound infection, recurrence, and prolonged period of recovery and improvement.

Table 49 Descriptive analysis of outcomes included in the Delphi study

Outcomes	Participant group	Round 1 N (%) 1-3	Round 1 N (%) 4-6	Round 1 N (%) 7-9	Round 2 N (%) 1-3	Round 2 N (%) 4-6	Round 2 N (%) 7-9	Category
To decrease the child's pain and muscle soreness								
To feel less pain	HCPs	4 (4)	9 (10)	80 (86)	1 (1)	9 (13)	58 (85)	A
	Patients/ Reps	2 (12)	3 (18)	12 (71)	2 (12)	1 (6)	14 (82)	
To feel less muscle soreness or tiredness after activities	HCPs	6 (6)	25 (27)	62 (67)	1 (1)	23 (34)	44 (65)	A*
	Patients/ Reps	3 (18)	5 (29)	9 (53)	-	3 (17)	15 (83)	
To improve the way the child looks								
To be able to stand taller or more upright	HCPs	7 (8)	30 (33)	55 (60)	3 (4)	21 (31)	44 (65)	C
	Patients/ Reps	2 (12)	4 (24)	11 (65)	4 (24)	2 (12)	11 (65)	
To have straighter and better shaped legs	HCPs	10 (11)	42 (46)	40 (43)	5 (7)	41 (60)	22 (32)	C
	Patients/ Reps	3 (17)	4 (22)	11 (61)	2 (11)	3 (17)	13 (72)	
To have legs that look the same as each other	HCPs	16 (17)	43 (46)	34 (37)	13 (19)	39 (57)	16 (24)	B*
	Patients/ Reps	5 (29)	1 (6)	11 (65)	4 (24)	3 (18)	10 (59)	
To increase how much the child can do								
To reduce or prevent falls	HCPs	1 (1)	15 (16)	77 (83)				A
	Patients/ Reps	1 (6)	-	17 (94)				
To be able to climb stairs	HCPs	5 (5)	34 (37)	53 (58)	1 (1)	31 (46)	36 (53)	C
	Patients/ Reps	2 (11)	5 (28)	11 (61)	2 (11)	5 (28)	11 (61)	
To be able to do more activities at home	HCPs	1 (1)	8 (9)	84 (90)	-	8 (12)	60 (88)	A
	Patients/ Reps	1 (6)	4 (22)	13 (72)	1 (6)	2 (11)	15 (83)	
To improve participation in sports (e.g., football, swimming)	HCPs	3 (3)	26 (28)	64 (69)	1 (1)	26 (38)	41 (60)	C
	Patients/ Reps	4 (22)	5 (28)	9 (50)	3 (17)	4 (22)	11 (61)	
To improve participation in hobbies (e.g., dancing, horse-riding)	HCPs	2 (2)	18 (19)	73 (78)	1 (1)	19 (28)	48 (71)	A**
	Patients/ Reps	2 (12)	5 (29)	10 (59)	3 (17)	3 (17)	12 (67)	
To be able to join more activities at school (e.g., PE) *	HCPs				-	12 (18)	56 (82)	A**
	Patients/ Reps				5 (28)	1 (6)	12 (67)	B*
To be able to stand with a better balance *	HCPs				-	9 (13)	59 (87)	A
	Patients/ Reps				2 (11)	-	16 (89)	
To increase the child's independence								
To be able to go out of the house independently (without help from an adult)	HCPs	3 (3)	8 (9)	82 (88)				A
	Patients/ Reps	2 (11)	1 (6)	15 (83)				
To be able to do things independently at home	HCPs	1 (1)	8 (9)	84 (90)				A
	Patients/ Reps	2 (11)	1 (6)	15 (83)				
To be able to do activities of daily living independently (e.g., bathing, dressing and self-care)	HCPs	1 (1)	8 (9)	84 (90)				A
	Patients/ Reps	2 (11)	1 (6)	15 (83)				
To be able to walk without assistance when going out	HCPs	2 (2)	22 (24)	67 (74)	2 (3)	15 (22)	51 (75)	A
	Patients/ Reps	4 (22)	4 (22)	10 (56)	2 (11)	0	16 (89)	
To be able to walk without assistance at home	HCPs	1 (1)	15 (16)	75 (82)				A
	Patients/ Reps	3 (18)	1 (6)	13 (76)				
To improve the way the child walks after surgery								
To be able to walk with straight legs	HCPs	3 (3)	41 (45)	48 (52)	1 (1)	24 (35)	43 (63)	A*
	Patients/ Reps	3 (17)	4 (22)	11 (61)	2 (11)	1 (6)	15 (83)	
To be able to walk without getting tired	HCPs	2 (2)	14 (15)	77 (83)				A
	Patients/ Reps	1(6)	3 (17)	14 (78)				
To be able to walk faster	HCPs	5 (5)	35 (38)	52 (57)	3 (4)	24 (35)	41 (60)	C

	Patients/ Reps	1 (6)	9 (50)	8 (44)	3 (17)	4 (22)	11 (61)	
To be able to walk further	HCPs	2 (2)	18 (19)	73 (78)				A
	Patients/ Reps	2 (11)	2 (11)	14 (78)				
To be able to walk without using a crutch or walker	HCPs	6 (7)	41 (45)	44 (48)	3 (4)	30 (45)	34 (51)	C
	Patients/ Reps	4 (22)	1 (6)	13 (72)	3 (17)	3 (17)	12 (67)	
To be able to walk better even if continue using a crutch or walker	HCPs	1 (1)	9 (10)	82 (89)	-	8 (12)	60 (88)	A**
	Patients/ Reps	4 (22)	2 (11)	12 (67)	3 (17)	4 (22)	11 (61)	
To stop wearing a brace /splint/ orthosis	HCPs	13 (14)	41 (44)	39 (42)	14 (21)	28 (42)	24 (36)	A*
	Patients/ Reps	4 (22)	4 (22)	10 (56)	3 (17)	1 (6)	14 (78)	
To be able to walk with the foot flat on the ground *	HCPs				2 (3)	24 (35)	42 (62)	A*
	Patients/ Reps				1 (6)	1 (6)	16 (89)	
To improve how the child feel when interacts with other people								
The child improves friendships and relationships with other people	HCPs	11 (12)	32 (34)	50 (54)	5 (7)	27 (40)	36 (53)	C
	Patients/ Reps	4 (22)	9 (50)	5 (28)	2 (11)	9 (50)	7 (39)	
To feel more confident	HCPs	5 (5)	20 (22)	67 (73)	3 (4)	9 (13)	56 (82)	A**
	Patients/ Reps	3 (17)	1 (6)	14 (78)	2 (11)	4 (22)	12 (67)	
To feel happier	HCPs	6 (6)	22 (24)	65 (70)	2 (3)	12 (18)	54 (79)	A**
	Patients/ Reps	3 (17)	3 (17)	12 (67)	1 (6)	4 (22)	13 (72)	
To feel better about how other people treat the child	HCPs	11 (12)	27 (30)	52 (58)	6 (9)	27 (40)	35 (51)	B*
	Patients/ Reps	6 (33)	3 (17)	9 (50)	4 (22)	5 (28)	9 (50)	
Concerns about having surgery								
Feeling pain after the surgery	HCPs	6 (6)	27 (29)	60 (65)	2 (3)	20 (29)	46 (68)	C
	Patients/ Reps	2 (11)	3 (17)	13 (72)	1 (6)	5 (28)	12 (67)	
Developing wound infection	HCPs	4 (4)	39 (42)	50 (54)	3 (4)	26 (38)	39 (57)	C
	Patients/ Reps	3(17)	4 (22)	11 (61)	4 (22)	9 (50)	5 (28)	
Losing independence	HCPs	3(3)	15 (16)	75 (81)	2 (3)	5 (7)	61 (90)	A**
	Patients/ Reps	4 (22)	6 (33)	8 (44)	3 (17)	4 (22)	11 (61)	
Needing to have another surgery	HCPs	2 (2)	35 (38)	55 (60)	5 (7)	21 (31)	42 (62)	C
	Patients/ Reps	1 (6)	6 (33)	11 (61)	2 (11)	8 (44)	8 (44)	
Getting worse after surgery	HCPs	3 (3)	11 (12)	78 (85)	2 (3)	5 (7)	61 (90)	A**
	Patients/ Reps	1 (6)	4 (22)	13 (72)	5 (28)	5 (28)	8 (44)	B*
Taking a long time to walk again	HCPs	3 (3)	25 (27)	65 (70)	2 (3)	21 (31)	44 (66)	C
	Patients/ Reps	2 (11)	4 (22)	12 (67)	3 (17)	6 (33)	9 (50)	
How important are the following factors in your decision to recommend the surgery?								
Good access to physiotherapy services locally	HCPs	1 (1)	10 (11)	82 (88)				A
	Patients/ Reps	1 (6)	1 (6)	16 (89)				
Good communication with other healthcare professionals	HCPs	2 (2)	10 (11)	81 (87)				A
	Patients/ Reps	-	2 (11)	16 (89)				
Being well informed about the operation and potential risks	HCPs	-	6 (6)	87 (94)				A
	Patients/ Reps	-	-	18 (100)				
Getting help from the right health professionals when needed	HCPs	-	6 (6)	87 (94)				A
	Patients/ Reps	-	2 (11)	16 (89)				
Getting support from family/friends	HCPs	-	8 (9)	85 (91)	1 (1)	12 (18)	55 (81)	A**
	Patients/ Reps	-	7 (39)	11 (61)	2 (11)	3 (17)	13 (72)	
Ability to adhere to home exercises *	HCPs				2 (3)	10 (15)	56 (82)	A
	Patients/ Reps				1 (6)	2 (11)	15 (83)	

HCPs: Healthcare professionals Rep: Representatives (parents) * New outcomes suggested during round one
A: Consensus in by both groups A*: Consensus in by patients/Reps A**: Consensus in by HCPs
B*: Consensus out by patients/Reps C: No consensus by both groups

9.3.5 Analysis by clinical characteristics

There was a strong consensus on outcomes that reached 'consensus in' for individuals with CP who had different levels of disability according to the GMFCS. In contrast, there was a relationship between the scores that participants gave the items and the age of the respondent (>18 years old). This was due to the dominance of adults in this study. With regard to the time since surgery, those who had not undergone surgery at the time of the survey placed greater emphasis on most of the outcomes than those who had already undergone the surgery; who categorised the majority as 'important and critical'. This was not surprising since the results of the qualitative interviews had suggested that the emphasis placed on some outcomes was greater among younger patients who had not yet undertaken the surgery who held strong views on what it might bring for them in the future.

9.3.6 Linkage outcomes to the ICF-CY

Each outcome that reached 'consensus in' was linked to the ICF-CY. Outcomes were linked to the most specific possible level of the framework. ICF-CY coding of the 27 outcomes resulted in 23 linkages (Table 50). Outcomes that were related to individuals with CP and their representatives spanned 18 ICF-CY components; however, the majority of linkages (55.5%) were to the activity's outcomes component (see Table 51). The remaining links were to the body function/structure component (33.3%) and environmental factors (11.1%). Some outcomes/elements of outcomes (n= 14) could not be linked to a corresponding ICF-CY code. These outcomes encompassed broad concepts related to quality of life, independence and adverse events.

Outcomes that reached consensus among health professionals were also coded to the ICF-CY. This process resulted in 19 links (see Table 51). Similar to the finding with the individuals with CP and representatives' group, the majority of linkages were found to occur for the activity and participation component (63.1%), body function/structure (15.7%) and environmental factors (21%). Again, the researchers could not link a small number of outcomes/elements of outcomes to a corresponding ICF-CY code. These outcomes were related to constructs such as quality of life, independence, and adverse events.

Table 50 Results of ICF-CY linkages for 'consensus in' items

ICF-CY Components	ICF-CY Code	Description
Body Function/ Structure	b280	Sensation of pain
	b4552	Fatiguability
	b1266	Confidence
	b2402	Sensation of falling
	b770	Gait pattern
	s750	Structure of lower extremity
Activity	d230	Daily life activities
	d530	Self-care: Toileting
	d540	Self-care: Dressing
	d2101	Undertake a complex task
	d4152	Maintain body position
	d4501	Walking long distance
	d4508	Walking: not specified (Endurance)
	d4600	Moving around within the home
	d4601	Moving around within community
	d465	Moving around using equipment
	d6504	Maintaining assistive devices
Participation	d9201	Recreation and leisure
	d9204	Recreation and leisure: hobbies
Environmental Factors	e310	Support: Family
	e320	Support: Friend
	e355	Support: Health professional
	e580	Health services, systems and policies

Table 51 Distribution of ICF-CY components links of all outcomes that reached consensus [n (%)]

	Health professionals	Individuals with CP/representatives
Body function/structure	3 (15.7)	6 (33.3)
Activity	10 (52.6)	10 (55.5)
Participation	2 (10.5)	0
Environmental factors	4 (21)	2 (11.1)
Total linkage	19	18

9.4 Discussion

Using the methodological guidance offered by initiatives such as COMET (10), we performed a Delphi study to provide an international, multi-stakeholder, consensus-base of outcome domains that could be used to assess the efficacy and effectiveness of lower limb orthopaedic surgery in ambulant children with CP in clinical trials. Following two rounds of the survey, 16 outcomes reached our pre-defined consensus threshold for inclusion in the future consensus meeting. Further outcomes reached consensus by individual panels, but these were not taken forward as they did not meet our previously defined and published consensus criteria (295). However, it was planned that they would be discussed during the consensus meeting with stakeholder groups.

The healthcare professionals who took part in our study showed less certainty regarding which outcomes were core compared with parents and parent representatives. This was shown by their rating more outcomes as critically important than the patients and representatives rated in this way. This may reflect the significant challenges for the professionals of selecting core outcomes in conditions that involve high levels of heterogeneity (i.e., functional levels, age groups). Clinical evidence suggests that children with CP are not expected to change but the manifestation of CP that is established over time is a result of natural maturation or development (508). Differences in experiences and knowledge may also have influenced several of the professionals' ability to judge the importance of outcome domains. Several health professionals reflected in their feedback on the survey that different outcomes were important at different times. It was assumed that, in contrast, patients and representatives scored the outcomes based on their experiences.

Several outcomes that seemed important in the interviews did not make it past the agreement threshold (75%). Pain and fatigue were repeatedly discussed by patients and their representatives. Although fatigue did not reach consensus overall, it did in the patients and representatives' group. The importance of lower limb symmetry and alignment varied with each round. It is not possible to conclude that this was due to an increase in consensus or to the withdrawal of panel members whose opinions were not in line with the consensus. In the second round, only 24% of health professionals ranked symmetry as critical compared with 59% of patients and representatives, and 32% of health professionals ranked alignment as critical against 72% of patients and representatives. Previous research into patient and family concerns and the problems they would like to discuss in clinic consultations found that these outcomes were important (144, 209). These findings likely reflect the greater emphasis that patients and

representatives place on outcomes related to activities and participations, than body structure. These outcomes did not reach the threshold of importance level; it seems that they were seen as important but not core.

Only two outcomes related to motor function gained consensus. These were associated with the ability to assess change over time in a patient's fall incidence and ability to maintain their standing balance. These outcomes were regarded as essential as they have an impact on mobility and a person's quality of life. More than 90% and 85% of both participant groups ranked fall avoidance and standing balance as critically important, respectively. Adults with CP experience a decline in mobility as they observe their balance to worsen and their risk of falling to increase in early to middle adulthood (509). Our findings confirmed this research as 40% of the patients undergone surgery were adults. It is well-recognised that there is a need for increased consistency and psychometric rigour in the evaluation of standing balance in CP populations in order to advance understanding and implementation of optimal interventions to improve mobility and reduce the incidence of falls (509, 510).

The impact of surgery on gait abnormality to increase the ability to walk plantargrade and with straight legs reached consensus amongst the patients and representatives, but not the health professionals. It is of note that these outcomes have been used as core measures of effectiveness in most primary studies and systematic reviews that have been performed to date. In the survey, the outcomes were described in lay terms for both panels. It could be that the health professionals viewed these differently to scientific measures, because they tend to use scientific terms as clinical and objective statements (kinetic and kinematic data) instead of proxy measures for the impact of a condition. However, many health professionals did assign scores for the measures as they were described, and only a few selected 'important but not critical', so this finding suggests that they did not consider the measures in a different light because simple language was used.

Life impact (long term) outcomes that related to mobility, including gait, daily life activities and participation, drew scores across consensus levels. Perhaps these measures were too generalised for health professionals to commit their achievement to a specific time, because other factors (e.g., GMFCS level) would influence this decision. This consideration makes it difficult for the researcher to interpret the scoring and feedback received. For instance, certain patients could be perceived to be less mobile preoperatively, which led health professionals to reach consensus on walking with the use of assistive

devices. It was assumed that professionals found it difficult to assign a score irrespective of preoperative status.

Five statements that were offered under independence in the life impact domain gained consensus (75% -90%) between the stakeholder groups, although some outcomes that were related to mobility were rated as not critical by health professionals. It should be noted that the independence outcomes were not related to decreased use of mobility devices but of external support. These outcomes are rarely measured in lower limb orthopaedic surgery studies. This suggests that the study has identified a gap between what was regarded as important by participants in the panels and what is measured in studies.

Health-related quality of life was rated lowest of these outcomes, even though outcomes related to quality of life would be expected to affect individuals with CP and their representatives in important ways. It was felt by the researchers that the description of quality of life measures in the survey questions was a challenge due to the subjectivity of this domain. This challenge was noted in interviews with stakeholders. The domain was not presented as 'quality of life' verbatim, but as 'to improve how the child feels when they interact with other people'. It is possible that this phrase was misinterpreted by this group of participants; however, if this was the case, the researchers would have expected comments on these outcomes when respondents were asked for additional outcomes of importance in the first round. Low ratings in this section may also have been due to overlap among these quality of life outcomes and others that may have been seen as interchangeable and which impact indirectly on a person's quality of life, such as independence and mobility level.

Concerns about possible adverse consequences of surgery did not reach consensus within the patients' and representatives' panel. This section was not named as 'adverse events', but as 'Concern about having surgery'. Two possibilities may have affected the results. Firstly, it is possible that different concerns had different effects upon patients' personal experiences of surgery. Secondly, it is recognised that these results could have been affected by the small sample size of the group, which could have led to an underestimation of the importance of these outcomes. The results suggest that the health professionals valued both positive and negative outcomes of the intervention. Although patients assigned low scores to these measures, we would recommend that adverse events of surgery are measured in clinical trials and routine clinical settings post-surgery to prove the safety profiles of the interventions.

Another key aspect of COS development is how contextual factors (i.e., potential confounders) are defined alongside core outcome domains (498). The contextual factors area reached a very high level of consensus across stakeholders. Research that is emerging highlights the importance of involving many different stakeholders in surgical decision-making and planning; however, it has received little attention in trials. Such involvements of different stakeholders would introduce consideration context, but it is difficult to measure context in clinical trials that are aimed to assess the efficacy of interventions, in which evaluation of such factors might be beyond the scope of the trials. Use of COSs counteracts this problem because the choice of core outcomes can reflect the most clinically relevant factors.

The young people and parent participants stressed that their decision-making and consequently their satisfaction with the outcome of surgery could be highly influenced by factors unrelated to the efficacy or effectiveness of the intervention itself. These factors included the level of communication between them and the professionals, the information resources made available to them, availability of health services locally and the child's ability to adhere to the treatment requirements. Therefore, we suggest that future research should include the evaluation of patient satisfaction with healthcare services alongside that of the effectiveness of outcome measures.

Representation of participants

Three areas may have affected the representativeness of the study findings. Firstly, the health professionals' panel contained larger proportions of surgeons and physiotherapists than of other healthcare professionals, meaning that the views of other health professionals may have been under-represented. However, this panel constitution is reflective of current lower limb management, which is usually carried out through surgery and post-surgical rehabilitation in most cases. Secondly, the number of children and young adults was smaller than that of the adults. Thirdly, there was a slightly greater proportion of participants who had CP and had long-term experience since their operations than would be expected within the general population. This reflects that adults with CP require life-long assistance and care and must navigate the long-term benefits and consequences of orthopaedic surgery. This higher proportion of adults with long-term experience of the effects of lower limb orthopaedic surgery may have skewed the priorities from the individuals with CP group towards the long-term outcomes of the intervention. During analysis of future data, researchers should pay greater attention to ensure variation among panel members, as one group may influence the final contents of a core set if outcome scores are simply combined across stakeholder group.

An effort was made to ensure that the outcome list for the Delphi survey was concise and that there were no duplications, as it has been seen in previous studies that a long list of outcomes is significantly associated with a low response rate (511). Even so, it was difficult to engage children who were being treated for lower limb musculoskeletal deformities, and this may have been due to three reasons, explained in Table 52.

Table 52 Reasons of low response rate from children with CP

Reasons	Description
The method of approach	- Researchers assumed that participants who had been recruited online were not as invested in the study as those who had been recruited by personal contact with their local care team. - Many patients had no ongoing or active involvement with medical services or charities due to the coronavirus pandemic, which spread rapidly worldwide during the performance of this research. - The development of a short video explanation by a lay person of the COS Delphi study was seen as an ideal way to draw in respondents as it established the reasons for and the importance of the Delphi survey.
Inclusion from multiple countries	- Inclusion of patients from multiple countries has been reported to be more difficult than the inclusion of health professionals from several countries (10). - This could be because the study was conducted in English, so it was more receptive to potential respondents in Western countries and North America. - A review of 227 COS studies reported that the majority involved collaborators (79%) and participants (68%) from Europe and North America (478).
Impact of social media	- Most of the health professionals who participated were recruited from professionals organisations, whereas most patients were recruited openly through social media or charity/society newsletter advertisements. - Gargon (2016) noted that social media followers may show a lack of diversity (512).

Attrition rate of participants

The overall response rate across the Delphi surveys was 59.4%. Consequently, the attrition rate between rounds was 40.5%. The response rate among health professionals (65.5%) was higher than that of the patients and representatives (27.7%) in round two.

Gene et al. (513) suggested that participants with minority opinions were more likely to drop out between rounds of Delphi surveys. However, the attrition rate in this study was higher than that observed in other COS studies. There could be a few reasons for this as explained in Table 53.

Table 53 Reasons of attrition rate

Reasons	Description
Familiarity with the study methods	- Health professionals may be more familiar with completing research-related prioritisation exercises than patients.
Recruitment approach (Social Media)	- Recruitment to the patients' group, which included children, was based on online social media. - A previous COS study noted that patients who were recruited through social media had a higher attrition rate than those recruited through a hospital site (514).
Recruitment approach (Snowballing approach)	- It was assumed that these initial invitations and encouragement to take part were sent by known contacts of these participants. - The invitation to take part in round two was sent by the study team. - The study team email may not have been recognised by participants or they may have felt less obliged to complete the survey the second time (515).
Emphasis on the two-round completion	- The importance of completing both rounds of the Delphi surveys may have not been emphasised enough through social media and charity advertisements. - It was mentioned in the information sheet that the survey comprises two rounds. - Several reminder emails were sent to all participants who had completed round one.
Times between rounds	- It was assumed that the attrition rate was probably increased because there was a longer-than-anticipated gap (six weeks) between the rounds. - This gap may have led participants to lose familiarity with the details of the study and the interest required to continue.
Minority views	- Our online software shows that few logged in to complete round two. - It would therefore seem that the way in which outcomes were presented, or the perception of holding minority views, contributed to their withdrawal.

Strengths and limitations

This study was developed using robust methodology in accordance with recommendations from the COMET initiative. The Delphi panels consisted of different sets of expert stakeholders and international contributors who were recruited through the use of a range of methods to create a diverse sample.

There were some limitations to this study. First, the list of outcomes for the Delphi survey was produced from a systematic literature review, which had been performed only in the English language. The qualitative interviews that informed the outcomes list were conducted through a sampling frame with stakeholders in a single tertiary centre. It is possible that further outcomes might have been collected if the systematic literature review had covered non-English language publications and/or the qualitative interviews had been done nationally or internationally rather than among a regional population. However, the first round of the Delphi survey enabled all international participants to suggest additional outcomes, which were considered by the study team for inclusion in the second round.

Second, a relatively small number of patients were involved in the Delphi rounds. Although there is no guidance on panel size, it is recognised that results may be sensitive to the small size of groups if consensus is defined by this group. As mentioned before, this may have been due to the recruitment strategy. Third, the Delphi surveys were only conducted in English language due to the limitation of time.

9.5 Conclusion

The results of this study highlight many outcomes that were judged as most important to various stakeholders, but which are currently poorly reported in lower limb orthopaedic surgery trials, such as the importance of capturing the longer-term impact of the surgery. The outcomes generated by this study may also provide insight into priority areas for further research.

Findings from this Delphi survey were taken forward and discussed regarding their importance and potential inclusion as part of a COS in a virtual consensus meeting that was held with international multi-stakeholder groups (Chapter 10).

Next step

The results of the two-round survey (Table 49) were taken forward and presented at a consensus meeting with key stakeholder groups to discuss the results and finalise the core outcome set. The meeting discussed the following outcome categories:

- The outcomes that reached consensus as important to be at the core outcome set by both stakeholder groups (n=16),
- The outcomes that reached consensus as important from one of the two stakeholder groups (n=8 from healthcare professionals, and n=4 from individuals with CP and their parents),
- The outcomes that reached consensus as not important to be in the core outcome set (n=4), and
- The outcomes that reached no consensus by the stakeholder groups (n=11).

Chapter 10 Consensus Meeting: Selection of Outcome Domains

A core outcome set for lower limb orthopaedic surgery for children with cerebral palsy: an international multi-stakeholder consensus meeting

In this chapter, the final stage is described in the development of a COS in terms of domains to be used in clinical trials of lower limb orthopaedic surgery for ambulant children with CP. This chapter details the international consensus meeting, at which the researchers sought to reduce the number of outcome domains to a minimum and hence to define the final core set. This chapter was adopted and extracted from a published paper (465).

10.1 Background

As described in the introduction and literature review section 3.3, it is important that the surgery must be agreed upon high-quality evidence. However, in the absence of uniform outcomes and measurements, verifiable data cannot be collected, nor accurate results aggregated and refined through systematic review and meta-analysis (254). To provide our profession with this important set of outcomes, an international consensus meeting was organised in which participants could standardise measures of success in lower limb orthopaedic surgery. The meeting aimed to draw up recommendations regarding ways to standardise surgical outcomes toward production of a COS for use in future studies in this field. This COS would reflect the views of experienced health professionals, researchers, and children and young people affected by the condition, and their family members.

The COS consensus meeting was informed by four studies (Table 54), which were explained in detail in the previous chapters. Thus, this research aimed to achieve consensus, at an international level, among children, families and health professionals on a core set of domains to be measured during lower limb orthopaedic surgery performed on ambulant children with CP.

Table 54 Four previous studies

Study	Description	Reference
Qualitative Evidence Synthesis (Chapter 6)	- Identified the aspects of health and outcomes that were considered important from the perspectives of ambulatory children with CP and their parents.	(307)
Scoping Review (Chapter 7)	- Identified reported outcomes in surgical trials	(227)
Qualitative study (Chapter 8)	- Investigated and explored outcomes that were considered important to stakeholders	(425, 516)
Delphi Process (Chapter 9)	- Prioritised surgical outcomes	(465)

Research question

The following question guided this consensus meeting: What surgical outcomes and domains should be considered in the COS?

10.2 Methods

Ethical approval for the consensus meeting was gained from the Oxford Research Ethical Review Committee, UK.

10.2.1 Study design

In previous COS studies, various methods have been used to conduct a consensus meeting. For example, in some studies, meetings were held with participants from a range of stakeholder groups (502, 517). In other studies, separate meetings have been conducted for each stakeholder group (518).

For this study, it was considered important to have a range of stakeholders present at the same meeting for several reasons: to ensure that outcomes in the final core set fairly represented their shared priorities; to enable each group to hear the others' views, which would facilitate open discussion; and to seek convergence of stakeholder views.

Due to travel restrictions that were related to the COVID-19 pandemic, the main meeting was held virtually via Zoom. Because panel members were in different time zones, the meeting was conducted over two days (7th and 14th May 2021).

10.2.2 Participants

The COMET initiative recommends that consensus should be sought from a group of international experts in specific health conditions, including health professionals, patients and representatives. In accordance with this guideline, this study includes the views of different stakeholder group as presented in Table 55.

Table 55 Stakeholder groups in the consensus meeting

Panel	Criteria
Experienced healthcare professionals	- Healthcare professionals who were involved at the time of the study in the provision of care for lower limb surgical interventions. - This group included surgeons, rehabilitation professionals and nurses
Experienced academics (researchers)	- As defined by their publication status the field of CP research
Core Outcome Set advisors	- Researchers with expertise in core outcome set development
Adults with CP	- Adults with CP, who had experienced lower limb orthopaedic surgery in childhood
Representatives	- Parents or carers of children with CP.

10.2.3Sample size

There is no guideline on the optimal sample size for a face-to-face consensus method. Studies have found that researchers who are willing to participate in the consensus panel are generally representative of their colleagues (519). Previous COS developers have reported a range of participant numbers for consensus meetings. For example, Harman et al. (494) met with 14 participants to develop a COS for children with cleft palate. Schmitt et al. (520) conducted a consensus meeting with 43 participants to draw up an eczema COS.

For this study, it was agreed that the target participation figure should be between 20 and 30 multi-stakeholder experts. A guideline of the outcome measures in a rheumatology (OMERACT) initiative suggests that at least 10% of the participants should be patients and representatives (299). Therefore, this study invited 6 adults with CP and/or their representatives to take part. This figure accounted for approximately 20% of the required total group of 20-30 experts.

10.2.4 Recruitment

It was intended that the consensus meeting would be internationally representatives, and hence participants were invited from four continents (Europe, North America, Australia and Asia), with the aim that at least one representative from each continent would take part. A formal email invitation was sent

to potential participants requesting their presence at the consensus meeting. The email included detailed information about the meeting. Potential panels were identified from existing contacts of members of the steering group (n= 8). Additional experts (n= 17) from six countries were identified through established relationships with the COS study team members, through requests to patients and health professionals' societies and from participants in the Delphi survey.

10.2.5 Ethical considerations

The researchers considered how they could ensure that patient and public representatives were well-informed and integrated into the discussion at the meeting. To aid understanding, all participants were provided with comprehensive information about the study in advance of the meeting. Patient and public participants were offered a pre-meeting discussion with the main researcher to ensure that they understood the purpose of the meeting, what was required of them, what they could expect to happen and how they could feel comfortable at the meeting.

At the start of the meeting, participants were encouraged to avoid jargon and to use language that would be acceptable to lay participants. Written consent was not obtained at the meeting as no identifiable data would be obtained during the proceedings. However, verbal consent was obtained to enable the researchers to audio-record the meeting. Discussion notes and the audio-recording were taken to inform the reporting of the results. Voting was anonymous.

10.2.6 Consensus process

This section describes the pre-meeting information that was circulated to participants, and the structure of the consensus meeting.

Pre-meeting information

The research team met monthly to ensure that the goals of the meeting were defined clearly and consistently and to prepare and review all the information that would be provided to the participants. All participants were provided with the following documents two weeks prior to the meeting: (1) the meeting agenda, (2) the background of the project, (3) an evidence synthesis of the methods that had been used to gather data before the consensus meeting, and (4) a summary of the Delphi findings, which provided the scores for each outcome.

Meeting structure

The consensus meeting structure comprised a combination of presentations of background information and data from previous stages, small- and large-group discussions and voting to achieve consensus. The key structure is described in the following section and Figure 23.

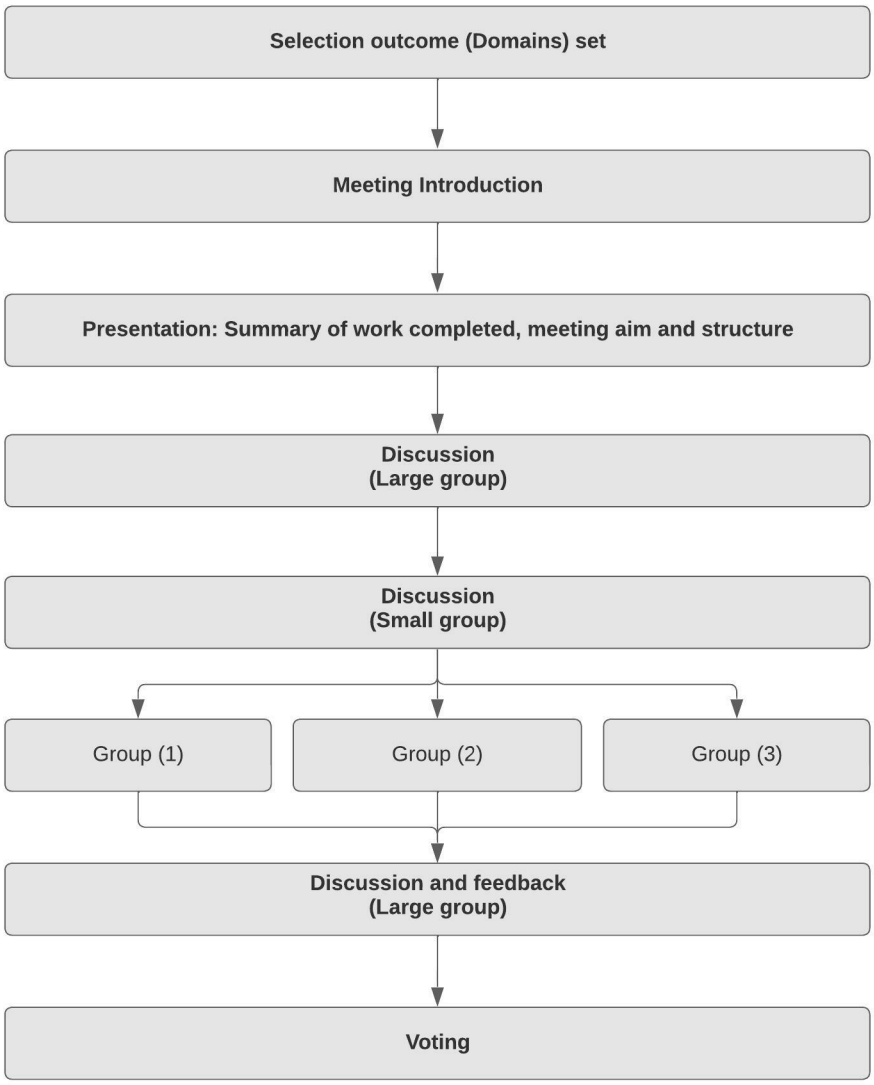


Figure 23 Consensus meeting agenda

Session 1: Introduction and background

The meeting was opened by stating the purpose of the meeting, which was to agree on core outcome and emphasised that the patient voice was crucial in determination of the COS and encouraged all members to engage with the discussion and share their views, knowledge and experiences.

A background of the project was then presented and explained, and the roadmap of the project was described to enable everyone to understand fully how we had defined the outcomes and domains, and the pre-meeting research findings were presented (the Qualitative Evidence Synthesis, the Scoping review and the interview study). This was followed by a presentation of the overview of the results of the Delphi survey, which revealed the number of outcomes given in each category that had reflected the agreement levels: 'consensus in', 'no consensus' and 'consensus out'. Finally, an overview of the meeting structure was explained (a large group discussion to reflect on the Delphi study findings would be followed by small group discussions, which would feedback to the large group before performance of interactive voting).

Session 2: Reflection on outcome domains

During the meeting, the outcomes that had been identified in the Delphi study as a whole were discussed. All outcomes were presented for each domain and each outcome was explained. Participants who considered that they were unable to judge an outcome, were able to indicate this. Outcomes identified as 'consensus in' were presented first. Participants gave feedback and reflections to the whole group on their opinions regarding which outcomes were the most important and why. Outcomes that had been deemed 'consensus out' were reviewed and participants were asked if they considered any fundamental reasons why these should be included in the COS. This was followed by a discussion of 'no consensus' outcomes, and similarly, participants were asked if they could think of any fundamental reasons why these should not be included in or dropped from the COS.

Session 3: Small group discussion

To ensure that all participants' views were considered, a breakout group discussion was then held. Participants were allocated a priori into three breakout groups to ensure representation in each group from the different stakeholder groups and countries. The purpose of this session was to discuss fully the outcomes that had been assigned to the 'no consensus' category. Facilitators who had been identified in advance of the meeting assisted in the small-group discussions. Facilitators had the role of encouraging participation of all group members, keeping discussions on topic and bringing together the key messages at the end of the group discussions. Each group was asked to consider the points in Table 56 in their discussions of each outcome.

Table 56 Small-group discussion

Points to consider in the small-group discussion
Whether there was an issue around the phrase used to describe the outcome (face validity)
Whether there was any conflict regarding the definition of the outcome. Participants were encouraged to reformulate outcomes to improve clarity and comprehension
Whether further factors should be considered before members of the meeting could judge the importance level of an outcome
Whether the outcome should be measured as a separate unique construct or whether it should be merged with one or more of the other outcomes
Whether the outcome should be moved from the 'no consensus' category to 'consensus in' or 'consensus out'

Session 4: Large group discussion

Once all groups had discussed the 'no consensus' outcomes, another large group discussion was organised. Facilitators and participants contributed to feedback key points and summaries from the small-group discussions.

Session 5: Voting and consensus development

Anonymised votes were taken on outcomes that had been categorised previously as 'no consensus' in order to distinguish their importance or non-importance. The votes allocated outcomes of sufficient importance to be included in the COS; important as optional outcomes, i.e., they might be impacted hugely by other factors or interventions; or not sufficiently important to be included in the COS. Participants voted online; votes were recorded with the use of Zoom. The results were presented to the group once the voting had closed. The study research team, who had facilitated the COS development process, did not participate in COS voting.

The meeting closed with a summary of agreed outcomes and the provisional list of those that would be included in the COS. Participants were asked to indicate whether they agreed or disagreed with this COS.

10.3 Results

10.3.1 Participants

Of 25 potential participants who were invited, four could not attend the meeting: a surgeon, a physiotherapist, a researcher, and an adult with CP. In total, 21 people participated in the consensus meetings that were held online (May 2021). These participants represented five countries: the UK, the USA, Canada, Australia and New Zealand. Panel members comprised: experienced clinicians and

researchers with expertise in the design and conduct of CP trials (n=15); adults with CP (n=3); and family members of a child with CP (n=3). Two adults with CP were from the UK and one from US; all were female. The representatives were parents of a children with CP; two were mothers and one was a father, who was a chair of a patients advocate society in the US.

10.3.2 Candidate domains

Combining outcomes from the initial stage of the project resulted in a list of 37 unique outcomes. Four additional outcomes were proposed during the first round of the Delphi survey. Results are summarised in Table 57 for each of the 41 outcomes. Based on the percentage scores, outcomes were categorised into each level of importance and organised into consensus categories A, B or C.

Table 57 Descriptive analysis of outcomes included in the Delphi study

Outcomes	Participant group	Round 1 N (%)			Round 2 N (%)			Category
		1-3	4-6	7-9	1-3	4-6	7-9	
To decrease the child's pain and muscle soreness								
To feel less pain	HCPs	4 (4)	9 (10)	80 (86)	1 (1)	9 (13)	58 (85)	A
	Patients/ Reps	2 (12)	3 (18)	12 (71)	2 (12)	1 (6)	14 (82)	
To feel less muscle soreness or tiredness after activities	HCPs	6 (6)	25 (27)	62 (67)	1 (1)	23 (34)	44 (65)	A*
	Patients/ Reps	3 (18)	5 (29)	9 (53)	-	3 (17)	15 (83)	
To improve the way the child looks								
To be able to stand taller or more upright	HCPs	7 (8)	30 (33)	55 (60)	3 (4)	21 (31)	44 (65)	C
	Patients/ Reps	2 (12)	4 (24)	11 (65)	4 (24)	2 (12)	11 (65)	
To have straighter and better shaped legs	HCPs	10 (11)	42 (46)	40 (43)	5 (7)	41 (60)	22 (32)	C
	Patients/ Reps	3 (17)	4 (22)	11 (61)	2 (11)	3 (17)	13 (72)	
To have legs that look the same as each other	HCPs	16 (17)	43 (46)	34 (37)	13 (19)	39 (57)	16 (24)	B*
	Patients/ Reps	5 (29)	1 (6)	11 (65)	4 (24)	3 (18)	10 (59)	
To increase how much the child can do								
To reduce or prevent falls	HCPs	1 (1)	15 (16)	77 (83)				A
	Patients/ Reps	1 (6)	-	17 (94)				
To be able to climb stairs	HCPs	5 (5)	34 (37)	53 (58)	1 (1)	31 (46)	36 (53)	C
	Patients/ Reps	2 (11)	5 (28)	11 (61)	2 (11)	5 (28)	11 (61)	
To be able to do more activities at home	HCPs	1 (1)	8 (9)	84 (90)	-	8 (12)	60 (88)	A
	Patients/ Reps	1 (6)	4 (22)	13 (72)	1 (6)	2 (11)	15 (83)	
To improve participation in sports (e.g., football, swimming)	HCPs	3 (3)	26 (28)	64 (69)	1 (1)	26 (38)	41 (60)	C
	Patients/ Reps	4 (22)	5 (28)	9 (50)	3 (17)	4 (22)	11 (61)	
To improve participation in hobbies (e.g., dancing, horse-riding)	HCPs	2 (2)	18 (19)	73 (78)	1 (1)	19 (28)	48 (71)	A**
	Patients/ Reps	2 (12)	5 (29)	10 (59)	3 (17)	3 (17)	12 (67)	
To be able to join more activities at school (e.g., PE) *	HCPs				-	12 (18)	56 (82)	A**
	Patients/ Reps				5 (28)	1 (6)	12 (67)	B*
To be able to stand with a better balance *	HCPs				-	9 (13)	59 (87)	A
	Patients/ Reps				2 (11)	-	16 (89)	

To increase the child's independence								
To be able to go out of the house independently (without help from an adult)	HCPs	3 (3)	8 (9)	82 (88)				A
	Patients/ Reps	2 (11)	1 (6)	15 (83)				
To be able to do things independently at home	HCPs	1 (1)	8 (9)	84 (90)				A
	Patients/ Reps	2 (11)	1 (6)	15 (83)				
To be able to do activities of daily living independently (e.g., bathing, dressing and self-care)	HCPs	1 (1)	8 (9)	84 (90)				A
	Patients/ Reps	2 (11)	1 (6)	15 (83)				
To be able to walk without assistance when going out	HCPs	2 (2)	22 (24)	67 (74)	2 (3)	15 (22)	51 (75)	A
	Patients/ Reps	4 (22)	4 (22)	10 (56)	2 (11)	0	16 (89)	
To be able to walk without assistance at home	HCPs	1 (1)	15 (16)	75 (82)				A
	Patients/ Reps	3 (18)	1 (6)	13 (76)				
To improve the way the child walks after surgery								
To be able to walk with straight legs	HCPs	3 (3)	41 (45)	48 (52)	1 (1)	24 (35)	43 (63)	A*
	Patients/ Reps	3 (17)	4 (22)	11 (61)	2 (11)	1 (6)	15 (83)	
To be able to walk without getting tired	HCPs	2 (2)	14 (15)	77 (83)				A
	Patients/ Reps	1(6)	3 (17)	14 (78)				
To be able to walk faster	HCPs	5 (5)	35 (38)	52 (57)	3 (4)	24 (35)	41 (60)	C
	Patients/ Reps	1 (6)	9 (50)	8 (44)	3 (17)	4 (22)	11 (61)	
To be able to walk further	HCPs	2 (2)	18 (19)	73 (78)				A
	Patients/ Reps	2 (11)	2 (11)	14 (78)				
To be able to walk without using a crutch or walker	HCPs	6 (7)	41 (45)	44 (48)	3 (4)	30 (45)	34 (51)	C
	Patients/ Reps	4 (22)	1 (6)	13 (72)	3 (17)	3 (17)	12 (67)	
To be able to walk better even if continue using a crutch or walker	HCPs	1 (1)	9 (10)	82 (89)	-	8 (12)	60 (88)	A**
	Patients/ Reps	4 (22)	2 (11)	12 (67)	3 (17)	4 (22)	11 (61)	
To stop wearing a brace /splint/ orthosis	HCPs	13 (14)	41 (44)	39 (42)	14 (21)	28 (42)	24 (36)	A*
	Patients/ Reps	4 (22)	4 (22)	10 (56)	3 (17)	1 (6)	14 (78)	
To be able to walk with the foot flat on the ground *	HCPs				2 (3)	24 (35)	42 (62)	A*
	Patients/ Reps				1 (6)	1 (6)	16 (89)	
To improve how the child feel when interacts with other people								
The child improves friendships and relationships with other people	HCPs	11 (12)	32 (34)	50 (54)	5 (7)	27 (40)	36 (53)	C
	Patients/ Reps	4 (22)	9 (50)	5 (28)	2 (11)	9 (50)	7 (39)	
To feel more confident	HCPs	5 (5)	20 (22)	67 (73)	3 (4)	9 (13)	56 (82)	A**
	Patients/ Reps	3 (17)	1 (6)	14 (78)	2 (11)	4 (22)	12 (67)	
To feel happier	HCPs	6 (6)	22 (24)	65 (70)	2 (3)	12 (18)	54 (79)	A**
	Patients/ Reps	3 (17)	3 (17)	12 (67)	1 (6)	4 (22)	13 (72)	
To feel better about how other people treat the child	HCPs	11 (12)	27 (30)	52 (58)	6 (9)	27 (40)	35 (51)	B*
	Patients/ Reps	6 (33)	3 (17)	9 (50)	4 (22)	5 (28)	9 (50)	
Concerns about having surgery								
Feeling pain after the surgery	HCPs	6 (6)	27 (29)	60 (65)	2 (3)	20 (29)	46 (68)	C
	Patients/ Reps	2 (11)	3 (17)	13 (72)	1 (6)	5 (28)	12 (67)	
Developing wound infection	HCPs	4 (4)	39 (42)	50 (54)	3 (4)	26 (38)	39 (57)	C
	Patients/ Reps	3(17)	4 (22)	11 (61)	4 (22)	9 (50)	5 (28)	
Losing independence	HCPs	3(3)	15 (16)	75 (81)	2 (3)	5 (7)	61 (90)	A**
	Patients/ Reps	4 (22)	6 (33)	8 (44)	3 (17)	4 (22)	11 (61)	
Needing to have another surgery	HCPs	2 (2)	35 (38)	55 (60)	5 (7)	21 (31)	42 (62)	C
	Patients/ Reps	1 (6)	6 (33)	11 (61)	2 (11)	8 (44)	8 (44)	
Getting worse after surgery	HCPs	3 (3)	11 (12)	78 (85)	2 (3)	5 (7)	61 (90)	A**
	Patients/ Reps	1 (6)	4 (22)	13 (72)	5 (28)	5 (28)	8 (44)	B*

Taking a long time to walk again	HCPs	3 (3)	25 (27)	65 (70)	2 (3)	21 (31)	44 (66)	C
	Patients/ Reps	2 (11)	4 (22)	12 (67)	3 (17)	6 (33)	9 (50)	
How important are the following factors in your decision to recommend the surgery?								
Good access to physiotherapy services locally	HCPs	1 (1)	10 (11)	82 (88)				A
	Patients/ Reps	1 (6)	1 (6)	16 (89)				
Good communication with other healthcare professionals	HCPs	2 (2)	10 (11)	81 (87)				A
	Patients/ Reps	-	2 (11)	16 (89)				
Being well informed about the operation and potential risks	HCPs	-	6 (6)	87 (94)				A
	Patients/ Reps	-	-	18 (100)				
Getting help from the right health professionals when needed	HCPs	-	6 (6)	87 (94)				A
	Patients/ Reps	-	2 (11)	16 (89)				
Getting support from family/friends	HCPs	-	8 (9)	85 (91)	1 (1)	12 (18)	55 (81)	A**
	Patients/ Reps	-	7 (39)	11 (61)	2 (11)	3 (17)	13 (72)	
Ability to adhere to home exercises *	HCPs				2 (3)	10 (15)	56 (82)	A
	Patients/ Reps				1 (6)	2 (11)	15 (83)	

HCPs: Healthcare professionals Rep: Representatives (parents) * New outcomes suggested during round one
A: Consensus in by both groups A*: Consensus in by patients/Reps A**: Consensus in by HCPs
B*: Consensus out by patients/Reps C: No consensus by both groups

10.3.3 Consensus meeting: voting

The results of voting for each outcome are shown in Table 58. During the discussion of the outcomes that did not reach the threshold for 'consensus in' during the Delphi study, six outcomes were considered important and were included in the final COS. The voting altered the consensus status of one surgical outcome from 'consensus in' to 'consensus out'; however, participants emphasised the importance for clinical decision-making.

Table 58 Voting results (%)

Outcome	Consensus in	Optional outcome	Consensus out
To improve the way the child looks			
To be able to stand taller or more upright	47%	33%	20%
To have straighter and better shaped legs	87%	13%	-
To increase how much the child can do			
To be able to climb stairs	67%	20%	13%
To improve participation in sports (e.g., football, swimming)	7%	60%	33%
To improve the way the child walks after surgery			
To be able to walk faster	29%	57%	14%
To be able to walk without using a crutch or walker	33%	53%	13%
To improve how the child feel when interacts with other people			
The child improves friendships and relationships with other people	7%	20%	73%
Concerns about having surgery			
Feeling pain after the surgery	20%	20%	60%
Developing wound infection	33%	40%	27%
Needing to have another surgery	40%	33%	27%
Taking a long time to walk again	40%	27%	33%

10.3.4 Consensus meeting: Panel discussion

During discussions, participants suggested that different aspects of outcomes might be important to different children, with the result that no individual outcome would reach the threshold for inclusion despite broad agreement that the outcome overall should be included. Participants regrouped or redefined some items from the list of 41 outcomes. For example, fatigue captures muscle fatigue and fatigue (or tiredness) after activities. Pain and fatigue were grouped under the larger concept of pain, discomfort and fatigue. Similarly, physical activity was considered to include both individual and groups sports. Specific details of each outcome discussion and their reformulation during the consensus meeting are provided in Appendices 11 and 12.

International Classification of Functioning- Children and Youth (ICF-CY): Participants discussed a wide range of outcomes that spanned across the ICF- CY framework. The group reached high level consensus in at least one outcome for each ICF-CY components including body function/structure and activity and participation, such as pain, fatigue, walking and physical activity. Discussion of outcomes related to the use of assistive devices caused some controversy; participants reached consensus that these should not be considered in the COS due to insufficient evidence to support the idea that the long-term impact of the surgery could decrease the use of assistive devices, particularly in children within GMFCS level III. Participants discussed the area of participation at length, due to the complexity of the domain and its reflection of individuals' own interest; thus, they concluded that 'physical activity' was a more appropriate termed to reflect the outcome across participation levels. While there was a general agreement not to discuss instruments, the use of an approach to assessment tool (e.g., the GOAL questionnaire) that was designed to capture the breadth of these domains was discussed briefly and was favoured.

Independence: Participants reached consensus and prioritised those outcomes which related to increased independence in various life roles. This is reflective of the agreement level in the Delphi study, in which all outcomes that were related to independence reached consensus in all groups. However, health professionals said that they were unclear of its contribution to the surgical intervention outcomes.

Quality of Life: The concept of quality of life was brought up for discussion since it potentially overlapped with some other domains. Participants agreed that aspects of physical and social life were included in other ICF-CY domains that could lead to improvement in the child's quality of life. However, participants

favoured the inclusion of specific outcomes in the core set (self-esteem and body image) over the broad concept of the ICF-CY framework.

Adverse Events (AEs): There was a debate regarding whether or not adverse events should be included as part of COS because their measurement was important in clinical trials. Discussions were focused on two main points: one of the main objectives of the surgery in CP is ultimately to prevent deterioration of musculoskeletal pathology and consequences; and it is difficult to demonstrate a significant difference in the occurrence of certain adverse events, such as infection, in long-term studies. The final decision was that adverse event reporting would not be a beneficial core outcome in trials of the effectiveness of lower limb surgical interventions, but an essential part of study documentation (Table 59). Several considerations that reflected this decision were discussed, including: the common understatement of both the benefits and the harm that surgery brings; the suggestion that such information would support patients and families during their decision making; the need to ensure collection of evidence of potential harm during the development of new surgical techniques; and the need to adhere to good clinical practice guidelines.

Factors impacting surgical decision-making: The participants reached a consensus that the factors that were listed in the survey that could affect the decision of whether or not to undertake surgery were all important. In addition, knowledge of these factors was essential to reach optimal surgical outcomes. However, it was decided that these factors were not always required to be reported in all clinical trials and therefore these factors should not be included as part of the COS. Table 60 shows the factors that were identified as important.

Table 59 Adverse event outcomes

Adverse Events
Post-Surgical Pain Syndrome
Wound infection
Losing independence
Recurrence
Getting worse than prior to surgery
Time to recovery

Table 60 Miscellaneous factors impacting decision-making

Adverse Events
Good access to physiotherapy services locally
Surgeon and Hospital factors, availability of good services
Being well informed about the operation and potential risks
Ability to adhere to home exercises
Getting support from family/friends

10.3.5 Core Outcome Set

At the end of the consensus meeting, eight domains were agreed on to be important and were proposed for inclusion in the final core outcome domain set for lower limb orthopaedic surgery for ambulant children with CP. These domains were: pain, discomfort and fatigue; lower limb structure; motor function; daily life activities; walking; physical activity; independence; and quality of life. The final COS is presented in Table 61.

Table 61 Core outcome set for lower limb surgery in children with CP

Domain (1) Pain, discomfort, and fatigue	**Domain (5) Mobility: Walking**
Pain	Walking pattern
Muscle fatigue	Walking distance
Fatigue (or tiredness after activities)	Maintenance walking speed
Domain (2) Lower limb structure	Walking endurance
Lower limb alignment	Ability to stop using orthosis, brace, splint
Lower limb symmetry	**Domain (6) Participation**
Domain (3) Motor function	Physical Activity
Standing position	**Domain (7) Independence**
Balance and fall	Daily life activities
Domain (4) Mobility: Daily Life Activity	Outdoor activities
Daily life activities (e.g., self-care, climb stair)	Walking without support from an adult
	Domain (8) Quality of life
	Self-esteem
	Body image

10.4 Discussion

Using robust methodology in accordance with the COMET initiative recommendations (10), we completed a meeting that produced an international, multi-stakeholder, consensus-based set of outcome domains that could be used in clinical research that aim to assess the efficacy or effectiveness of lower limb orthopaedic surgery in ambulant children with CP. The standardised reporting of core

outcomes that should be identified across future studies will help to improve the quality of evidence synthesis.

The consensus meeting derived the final COS, which consisted of 19 outcomes within eight domains. It is possible that the length of this list reflects the significant challenges of selecting core outcomes in conditions of high levels of heterogeneity (i.e., functionals levels, age group of affected children) and that different outcomes are important at different times. Clinical evidence suggests children with CP are expected to change that is established over time as a result of natural maturation or development (508). Furthermore, differences in experiences and knowledge are likely to influence each individual's ability to inform a judgement on the importance of each outcome domain.

Concern regarding potential adverse events of surgery did not reach consensus during the meeting for inclusion in the COS. Health professionals stated that they valued both positive and negative outcomes of surgical interventions. However, it is recommended that adverse events of surgery should be reported in future clinical studies to prove the safety profile of the interventions. Therefore, the adverse events domain was recommended for future study documentation.

Participants were advised to pay little attention to contextual factors during the consensus meeting, since evaluation of these factors might be outside the scope of research studies that would use the COS to measure the intervention efficacy or effectiveness. However, contextual factors have great prospective importance in the provision of care. This research has further highlighted the importance of discussing relevant factors during shared decision-making (e.g., communication, information resources). Consideration and management of these factors are likely to enhance the stakeholders' experiences postoperatively; therefore, a holistic evaluation of the process of treatment is recommended. Few studies have aimed to enhance or understand factors that are related to the provision of care in this field, so highlighting the need for further research in this area.

Strength and limitations

One strength of this study was that the findings of the meeting were further supported by the extensive evidence synthesis that was described in previous chapters and which was useful to inform this meeting. A further strength was that the views of both stakeholder groups were represented equally, despite differences in the numbers of panels in each group that took part in an in-depth discussion at the meeting. Some members of the healthcare professionals' panel were known international experts in the

field of lower limb orthopaedic surgery and CP. In addition, the inclusion of both patients and healthcare professionals at every stage enabled each group to hear the views of the other, which facilitated open discussion and ensured that outcomes in the final core set represented their shared priorities fairly.

There are some limitations to this study: for example, only English-speaking international experts could take part in the consensus meeting. It is possible that further outcomes may have been collected if the study had covered non-English speakers. To mitigate this, a wide range of outcomes was defined from worldwide reviews to inform the Delphi surveys (227, 307). In these reviews, the impact of language should not have been a major issue.

A further limitation of the study was that international representation of expertise was not attainable. Different countries have different healthcare systems and recovery experiences. Although great efforts were made to introduce our study to experts from different international backgrounds, participation was a challenge for some. This may have been because the meeting was conducted online and time differences between countries may have led to difficulties in taking part.

10.5 Conclusion

This COS was developed for studies and trials of lower limb orthopaedic surgery for ambulant children with CP. The COS included eight domains: pain and fatigue; lower limb structure; motor function; mobility; gait-related outcomes; physical activity; independence; and quality of life. The COS should underpin all future research studies. Implementation of the COS will standardise the selection, collection and reporting of outcomes, which ultimately will facilitate meta-analysis to inform clinical practice.

Next Step

The proposed COS will inform the next chapter, in which the development of a core outcome measurement set for each identified outcome/domain is described.

Chapter 11 Selection of Outcome Measures

Recommendation of core outcome measurements for ambulant children with cerebral palsy post lower limb orthopaedic surgery: a consensus meeting

In this chapter, recommendations for a core outcome measurements set for use in clinical trials of lower limb orthopaedic surgery for ambulant children with CP are described. This addressed the second objective strand to the research in this thesis, which provides recommendations on the best outcome measures to use to assess each of the core outcome domains identified in the previous chapter. This chapter brings both the quality level of current outcome measures and the feasibility of their measurement to an international consensus meeting. The meeting sought to reduce the number of outcome measures and hence to define the final set of core outcome measures. The results of this chapter have been drafted and the intention is to submit a paper to the journal 'Developmental Medicine and Child Neurology'.

11.1 Background

As described earlier, outcome measures must be valid, reliable, and responsive to change, otherwise, there is a serious risk of the generation of imprecise or biased results. In the field of lower limb orthopaedic surgery for ambulant children with CP, multiple outcome measures are available to measure the same health construct (outcome domains) (6, 227). The inconsistency and variability of outcome measures could hamper the comparability of results and increase the difficulty of carrying out a meta-analysis (6, 227, 295). As proposed by the COMET initiative, the development of a COS could address the problems that arise from outcome measure inconsistency (10, 215). The development of a COS is a two-step process: the first step is to determine which outcome domains should be included within the COS, and the second is to achieve consensus on how these outcomes could be measured (10, 256). For the purpose of the first step, a consensus was achieved on 19 outcomes within eight domains: pain and fatigue, lower limb structure, motor function, mobility (daily life activities), gait-related outcomes, ability to engage in physical activity, independence, and quality of life (presented in Chapter 10).

Thus, the aims of this study are (1) to identify the existing outcome measures that are used to assess the lower limb orthopaedic surgery outcomes; (2) to summarise and assess the current state of the quality and feasibility of the most frequently used outcome measures in the field of lower limb orthopaedic surgery for ambulant children with CP; and (3) to formulate consensus-based recommendations on core

outcome measurement instruments set for clinical studies in this field. The study focused mainly on the outcome measures for the eight core outcome domains set.

Research question

The research question for this study was: What are the most optimal outcome measurement tools that could be considered for evaluation of outcomes within the COS?

11.2 Methods

The COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) initiative (http://www.cosmin.nl/) aims to improve the selection of outcome measurement instruments and support the second step of development of a COS, which involves discovery of how to measure the outcomes. The research methods were based on guidelines that were developed and published by the COMET and COSMIN in 2016 (274).

The COSMIN checklist was chosen as the quality review tool because it involves multilevel grading of each measurement property within a standardised rating system and includes extensive guidelines that accompany the checklist (521). The COSMIN provides detailed standards specifically for the preferred design characteristics and statistical methods of studies that assess measurement properties (522). The checklist gives a grading for each measurement property, not the outcome measure as a whole. The COSMIN guideline was developed through Delphi studies in which a consensus was reached regarding the definitions and taxonomy, assessment of measurement properties and methods of selection of measurement tools for an agreed core outcome (domains) set (11, 220, 317, 522-525).

The selection of core outcome measurement instruments comprised the following steps: (1) Conceptual considerations; (2) Finding existing outcome measurement instruments; (3) Quality assessment of outcome measurement instruments; and (4) Generic recommendations on the selection of outcome measurement instruments for a COS.

Step one: Conceptual considerations

This step refers to the target population and construct (i.e., outcome or domain) to be measured. The target population of the proposed core outcome measurements set is associated with the proposed COS. Accordingly, in this study, available outcome measures that are used in clinical research following

lower limb orthopaedic surgery in ambulant children with CP and matching of the outcomes within the core outcome domains were considered for the core outcome measurements set.

Step two: Finding existing outcome measurement instruments

Numerous validated outcome measures for CP were identified from up-to-date scoping reviews (6, 191, 227). Chapter 7 detailed the methods of updating a scoping review of outcome measures that are used in the field of lower limb orthopaedic surgery for ambulant children with CP.

In summary, a systematic search was performed as described in Chapter 7. The PubMed, CINAHL, MEDLINE, EMBASE and Cochrane-controlled trial registry databases were searched from their inceptions until July 2019. Inclusion criteria required that the primary studies were focused on the reporting outcomes of lower limb orthopaedic surgery for ambulant children with CP (i.e., within Gross Motor Functional Classification System; GMFCS I-III).

The breadth and depth of the outcome measures content were examined and linked to the domains of the ICF-CY framework, independence and quality of life to support decisions regarding the suitability of the measures for the core domains. According to pre-specified criteria (Table 62), which were applied to the list of identified outcome measures, an initial screening was performed to determine whether each outcome measure matched the target outcomes or domains within the COS and to enable an initial reduction of the number of outcome measures.

Table 62 Criteria for initial reduction of the number of outcome measurement instruments

Measures were excluded if:
1. The purpose of the measurement instrument was to screen/set a goal for an intervention, rather than to measure outcomes
2. The measurement instrument was published more than 30 years ago (i.e., prior to 1990) without subsequent revision and/or was not in current use
3. Part of other measures

Step 3: Quality assessment of outcome measurement instruments

In accordance with the COSMIN guideline (274), judgement of the quality of an outcome measure required the evaluation of the evidence regarding the measurement's psychometric properties. Nine measurement properties were defined and subdivided into three major groups. These were: *validity:* the degree to which an instrument measured the construct(s) it purported to measure; *reliability:* the

degree to which the measurement was free from measurement error; and *responsiveness:* the ability of an instrument to detect change over time in the construct to be measured (Table 63) (274, 317).

Table 63 Measurement properties and their definitions

Term		Definition
Domain	**Measurement property**	
Validity	Content validity (including face validity)	The degree to which the content of a measurement instrument is an adequate reflection of the construct to be measured
	Structure validity	The degree to which the scores of a measurement instrument are an adequate reflection of the dimensionality of the construct to be measured
	Construct validity- hypothesis testing	The degree to which the scores of a measurement instrument are consistent with hypotheses based on the assumption that the measurement instrument validly measures the construct to be measured
	Cross-cultural validity	The degree to which the performance of the items on a translated or culturally adapted measurement instrument are an adequate reflection of the performance of the items of the original version of the measurement instrument
	Criterion validity	The degree to which the scores of a measurement instrument are an adequate reflection of a 'gold standard'
Reliability	Reliability	The proportion of the total variance in the measurements which is because of 'true'* differences among patients
	Measurement error	The systematic and random error of a patient's score that is not attributed to true changes in the construct to be measured
	Internal consistency	The degree of the interrelatedness among the items
Responsiveness	Responsiveness	The ability of a measurement instrument to detect change over time in the construct to be measured

* 'True' should be interpreted in the context of classical test theory, which states that any observation is composed of two components: a true score and an error associated with the observation. 'True' is the average score that would be obtained if the scale was given an infinite number of items, and it refers only to the consistency of the score and not to its accuracy.

The COSMIN initiative has led to the development of (1) Methodological quality analysis: methods to rate the methodological quality of studies with regard to the nine measurement properties; (2) Level of evidence synthesis: criteria by which to judge whether the results for each measurement property are satisfactory and (3) Best evidence synthesis: a system to rate the quality of the body of evidence on each property. Details of each quality assessment element are presented in the following sections.

Methodological quality analysis: The COSMIN initiative has led to the development of methods to rate the methodological quality of studies with regard to the nine measurement properties. It employs a four-point scoring system of excellent, good, fair and poor to provide an overall score for the methodological quality of a study for each category of measurement property under examination (see Appendix 13)(522). A different number of items is considered for each psychometric property (Table 64). For this study, in line with the COSMIN guideline, the overall rating of each category was given as the lowest level response to any item within that category (psychometric property).

Table 64 COSMIN checklist

COSMIN Checklist	Number of items
Internal consistency	11
Reliability	14
Measurement error	11
Content validity	5
Structural validity	7
Hypothesis testing	10
Cross-cultural validity	15
Criterion validity	7
Responsiveness	18

Level of evidence synthesis: The strength of the results within each study was also examined using published criteria, which are shown in Table 65 (526). The three possible levels of evidence are shown in this checklist. They are positive (+), indeterminate (?) and negative (-). Where no information is available, a zero (0) rating is given.

Table 65 Criteria for good measurement properties

Term		Rating	Definition
Domain	**Measurement property**		
Validity	Content validity (Including face validity)	+	All items refer to relevant aspects of the construct to be measured AND are relevant for the target population AND are relevant for the context of use AND together comprehensively reflect the construct to be measured
		?	Not all information for '+' reported
		–	Criteria for '+' not met
	Structure validity	+	Factors should explain at least 50% of the variance
		?	Explained variance not mentioned
		–	Criteria for '+' not met
	Construct validity hypothesis testing	+	At least 75% of the results are in accordance with the hypotheses
		?	No correlations with instrument(s) measuring related construct(s) AND no differences between relevant groups reported
		–	Criteria for '+' not met
	Cross-cultural validity	+	No important differences found between language versions in multiple group factor analysis or DIF analysis
		?	Multiple group factor analysis AND DIF analysis not performed
		–	One or more criteria for '+' not met
	Criterion validity	+	Convincing arguments that gold standard is "gold" AND correlation with gold standard ≥ 0.70
		?	Not all information for '+' reported
		–	Criteria for '+' not met
Reliability	Reliability	+	ICC or weighted Kappa ≥ 0.70 or Pearson's r≥ 0.80
		?	ICC or weighted Kappa not reported
		–	Criteria for '+' not met
	Measurement error	+	SDC or LoA < MIC
		?	MIC not defined
		–	Criteria for '+' not met
	Internal consistency	+	Cronbach's alpha(s) ≥ 0.70 and ≤ 0.95
		?	Cronbach's alpha not determined or dimensionally unknown
		–	Criteria for '+' not met
Responsiveness	Responsiveness	+	At least 75% of the results are in accordance with the hypotheses
		?	No correlations with changes in instrument(s) measuring related construct(s) AND no differences between changes in relevant groups reported
		–	Criteria for '+' not met

ICC = intraclass correlation coefficient, LoA = limits of agreement, MIC = minimal important change, SDC = smallest detectable change
* "+" = positive rating, "?" = indeterminate rating,"– "= negative rating

Best evidence synthesis: Researchers involved in the COSMIN initiative have also developed a system to rate the quality of the body of evidence for each property (Table 66) (274). To examine the best evidence synthesis, the methodological quality rating for the measurement properties of each outcome measure were presented alongside the level of evidence in each study. This enabled the drawing of inferences on the relative robustness of evidence for each outcome measure.

Table 66 Quality of evidence

Quality Rating	Criteria
Strong (+++)	Consistent findings in multiple studies of at least good quality OR one study of excellent quality AND a total sample size of ≥100 patients
Moderate (++)	Consistent findings in multiple studies of at least good quality OR consistent findings in multiple studies of at least fair quality OR one study of good quality AND a total sample size of ≥50 patients
Limited (+ or -)	Limited findings in multiple studies of at least fair quality OR one study of fair quality AND a total sample size of ≥30 patients
Conflicting (±)	Conflicting findings
Poor (?)	Only studies of poor quality OR a total sample size of <30 patients
Unknown (0)	No studies

COSMIN Database: The COSMIN database is freely accessible and contains systematic reviews of outcome measurement instruments for various health conditions (http://database. cosmin.nl/). For this work, the database was searched in July 2020 for studies that had investigated the clinimetric properties of outcome measures in children with CP in order to determine the current state of the quality of the identified outcome measures in the field of CP lower limb orthopaedic surgery. Existing systematic reviews were identified (n= 9), and the results were extracted and summarised. For example, Zanudin et al.(219) conducted a systematic review to assess gait-related outcome measures in CP through the use of a modified COSMIN method to choose suitable, high-quality outcome measures. Himuro et al. (527) systematically reviewed quick, simple and easy-to-use measures of walking ability that have been used to assess children with CP and evaluated the clinical utility and psychometric properties of these measures through the use of the COSMIN checklist.

Search source: A further search was conducted on July 2020 through the sensitive PubMed search filter, as proposed by COSMIN, for measurement properties used (528) for outcome measures that had no existing systematic review on their psychometric properties. Search key terms were: 1) general names for instruments; 2) multidimensional health construct terms; 3) cerebral palsy; and 4) key terms relevant to psychometric performance.

Feasibility criteria: The feasibility aspect of each outcome measure was also considered since it played an important role in the selection of outcome measures. This step required an evaluation of the interpretability, ease of administration, type and length of the outcome measure, its completion time, costs, the equipment required to use it, type of administration and other practical aspects.

The feasibility criteria of each outcome measure were identified from several resources such as the outcome measure's manual, primary development studies or existing reviews. For example, the CHQ and PedsQL characteristics were extracted from the review by Janssens et al. (287), which described PROMS that have been used for children with disabilities.

Data extraction: Data from the COSMIN database and the PubMed search were extracted through the use of standardised, piloted data extraction in Microsoft Excel. For each included outcome measure, the following were extracted: name, abbreviation, the purpose of measurement, number of items, age range, the responder (clinician, child and/or parent as proxy), response options, completion time, health domains assessed, methods for scoring, training requirement, evidence of COSMIN, best evidence synthesis criteria for the nine psychometric properties, and the key reference article.

Step four: Generic recommendations: Consensus meeting

The evidence that was generated in the second and third steps informed a consensus process to decide whether or not the individual outcome measures were recommended for the assessment of the core outcome domain. Ethical approval for the consensus meeting was gained from the Oxford Research Ethics Committee (19/SC/0357).

Five healthcare professionals from our project steering committee were invited to participate in an online consensus meeting via the Zoom platform on 27th September 2021. Two additional researchers from the US with expertise in CP were also invited to take part. Panel members were experienced in the design and conduct of CP research, measurement and testing.

Pre-meeting information

Monthly meetings were held among members of the research team to ensure clarity and consistency in the definition of the goals for the meeting and to prepare and review all the information that would be provided to the participants. To enable the invitees to participate fully in the meeting, they were provided with the following documents two weeks before it: (1) the meeting agenda; (2) background information regarding the core outcome measures set, including relevant details of the core outcome domains set; and (3) the current state of the outcome measures, in terms of their psychometric properties' status and feasibility criteria.

Meeting structure

The consensus meeting involved a combination of presentations of background information and data from previous stages, and whole group discussion to achieve consensus. The key points of the meeting are described in the following sections.

Session 1: Introduction and background

The meeting was opened by stating the purpose of the meeting, which was to agree on core outcome measurements. The group were encouraged to engage in the discussions and share their views, knowledge and experiences. This was followed by a presentation of some background of the project, which was explained to enable participants to understand fully and participate in the subsequent discussion. The roadmap was described to enable participants to understand fully how we had defined the outcome measures. This was followed by a presentation of the overview of the current state of the shortlisted outcome measures in terms of their quality and feasibility. Finally, the meeting structure was described so that participants knew the format; this was to take the form of a whole group discussion to reflect on the findings followed by a consensus discussion to finalise the core outcome measurements set.

Session 2: Reflection on outcome measures

At the meeting, the outcome measures for each of the eight domains were discussed. Participants who considered that they were unable to judge an outcome measure were able to indicate this. Participants engaged in a whole-group discussion and gave their feedback and opinions regarding the most optimal outcome measures, with consideration of these measures' suitability to match the outcome domains, psychometric properties and feasibility criteria. Outcome measures that were deemed not feasible were discussed and participants were asked whether they thought there were any fundamental reasons why these could not be included in the core set.

Once the panel had discussed all outcome measures for each outcome domain, the group contributed feedback on key points and the discussion was summarised. The meeting closed with a synopsis of the agreed outcome measures and the provisional list of the core outcome measurements set.

11.3 Results

11.3.1 Outcome Measure Instruments

Overall, 30 different outcome measures were identified from the updated review in the 44 studies. One of them, Dogan's Scale (529), was excluded as its psychometric properties have not been investigated. Thus, 29 outcome measures were included (Table 67). Outcome measures were categorised into the domains of body function and structure, activity and participation and adverse events and harm.

There were 23 clinician-administered measures (79% of the instruments), and six self/proxy administered measures. These were: the Pediatric Orthopaedic Data Collection Instrument (PODCI) (530); the Patient-Reported Outcome Measures Information System (PROMIS) (531); the Gillette Functional Assessment Questionnaire (Gillette FAQ) (183); the Pediatric Evaluation of Disability Inventory (PEDI) (304); the Computerised Adaptive Tests (CAT) (532); and the Mobility Questionnaire 47 (MobQues47) (533).

Twenty (69%) of the outcome measures that were included in the review applied to the measurement of impairment of body structure and function. Outcome measures that assessed gait pathology were the most common (37/44 studies, 84%) and included gait analysis and its parameters: kinematics, kinetics, Gait Profile Score (GPS), Gait Deviation Index (GDI) and Gillette Gait Index (GGI). Clinical examination was the second most common outcome measure that was assessed and included: data for the joint range of motion (ROM) that were captured by a goniometer (n=17, 39% of studies); muscle strength, which was measured through the use of manual muscle testing (n=6, 14% of studies); and muscle spasticity, which was measured through the use of the Modified Ashworth Scale (MAS) (n=5, 11%), the Tardieu Scale (n=2, 5%) and Duncan Ely's sign (n=1, 2%). Pain was only measured in four studies, as part of multi-dimension outcome measures such as PODCI, PROMIS and CAT.

The patient and proxy (parent) measures, which were most commonly administered by questionnaires or interviews with health professionals, were those used to assess the impact of surgery on activity and participation levels such as walking, mobility, social function and level of independence. There was inconsistency in the choice of measures: the Gillette FAQ and PODCI were each used in three studies (7% each), whilst PROMIS, MobQues47, CAT and PEDI were equally represented within the studies at n=1 (2%). The Functional Mobility Scale (FMS) (n=1, 2%) and the GMFM (n=2, 5%) were the only clinician-

administered tools that were used to assess motor function outcomes. Surgical adverse events (complications) and harm were measured using the Clavien-Dindo system (n=3, 7%).

Table 67 Outcome measure across 44 studies

Outcome Measures	Identified papers (n= 44)		Reference
	Number of papers	Percentage (%)	
Body Function and Structure			
Gait Analysis (Kinematics or Kinetics)	37	84%	(164-168, 383-386, 388, 390-414)
Gait Deviation Index (GDI)	12	27%	(164-166, 168, 392-394, 403, 404, 412-414)
Gillette Gait Index (GGI)	1	2%	(402)
Gait Profile Score (GPS)	8	18%	(384, 386, 392, 395, 397, 398, 406, 410)
Gait cycle	1	2%	(387)
Gait velocity	8	18%	(385, 387, 400, 402, 406-409)
Movement Analysis profile (MAP)	3	7%	(384, 397, 410)
Gait velocity Scale (GVS)	1	2%	(406)
Stahelil's Rotational Profile	1	2%	(396)
Radiology	7	16%	(385, 389, 399, 416, 417, 419, 420)
Foot pressure data	3	7%	(385, 417, 420)
Foot progression angle	3	7%	(166, 167, 404)
Range of Motion	17	39%	(164, 165, 386, 387, 390, 391, 397-399, 402, 404, 405, 407-409, 417, 418)
Modified Ashworth Scale (MAS)	5	11%	(165, 386, 397, 405, 417)
Tardieu scale	2	5%	(165, 398)
Duncan Ely's Sign	1	2%	(398)
Manual Muscle Test (MMT)	6	14%	(165, 386, 387, 397, 398, 421)
Oxygen consumption	2	5%	(166, 400)
Edinburgh Visual Gait Score (EVGS)	2	5%	(389, 407)
Timed-up and go	1	2%	(415)
Activity and Participation			
Gross Motor Function measures (GMFM)	2	5%	(164, 415)
Functional Mobility Scale (FMS)	1	2%	(421)
Gillette Functional Assessment Questionnaire (Gillette FAQ)	3	7%	(387, 415, 417)
Pediatric Evaluation of Disability Inventory (PEDI)	1	2%	(421)
Computerised Adaptive Tests (CAT)	1	2%	(415)
Mobility Questionnaire 47 (MobQues47)	1	2%	(421)

Multi-Dimensional Outcome Measure			
Pediatric Orthopaedic Data Collection Instrument (PODCI)	3	7%	(165, 166, 415)
Patient Reported Outcome Measures Information System (PROMIS)	1	2%	(164)
Adverse event and harm			
Clavien-Dindo System	3	7%	(395, 398, 410)
Total	137		

11.3.2 Comparison of the literature before and after 2016

A comparison of pre- versus post-2016 literature was based on the outcome measures, as outcome domains had not been identified in the original review. The number of outcome measures that were used decreased slightly from 34 before 2016 to 29 after 2016 (Table 68). As in the original review, the study designs were mostly retrospective and body structure and function was the most frequently measured outcome.

Nineteen of the measurement tools that had been included in the previous reviews (6, 191) were not identified in the updated review and nine additional measurement tools were identified that had not been included in the previous review (Table 68).

Although the number of outcome measures that were used to assess the impact of lower limb surgery remained stable between reviews, there were differences in the type and frequency of use of the outcome measures; for example, the GGI was identified in studies that were published both before and after 2016: however, the frequency was varied, (n=20, 6% and n=1, 2% respectively (Table 69)). In the updated review, QoL was measured through the use of multi-dimensional patient-reported outcome measures (PROMs) such as PROMIS, rather than through the use of disease-specific outcome measures such as CP-QoL (196) or the Paediatric and Quality of Life Inventory (PedsQL) (534) .

The GMFCS has been used in recent studies to describe study participants in terms of motor function level. Compared with pre-2016 findings, use of the GMFCS increased from 39% of the identified studies since its origin, between 2003 and 2016, and 84% over the 2016-2019 period.

Table 68 Outcome measures used only before or only after 2016. Please note that although the Normalcy Index is reported separately here, it is the same index that became known as the GGI shown in Table 69. The Normalcy Index appears on this Table as it was used under this name only before 2016.

Outcome measures	**Pre- 2016** (6, 191) N = 310	**Post- 2016** (227) N = 44
Body Function and Structure		
Type of walking device	29	-
Surface electromyography (EMG)	20	-
Presence of pain	14	-
Biomechanical Model	7	-
Physiological Cost Index	4	-
Normalcy Index (previous name of GGI)	3	-
Physician Rating Scale (PRS)	2	-
Hip Flexor Index	2	-
Observational gait (OG)	2	-
Vertical plantar pressure	1	-
Selective Control Assessment of the Lower Extremity	1	-
Total mechanical work	2	-
Movement Analysis Profile (MAP)	-	3
Gait Velocity Scale (GVS)	-	1
Stahelil's Rotational Profile	-	1
Foot progression angle	-	1
Edinburgh Visual Gait Score (EVGS)	-	2
Activity and Participation		
Functional Independence Measure for Children (FIM)	4	-
Positional Activity Logger (PAL)	1	-
Modified Goal Attainment Scale (Modified GAS)	1	-
Gross Motor Performance Measure (GMPM)	1	-
Computerised Adaptive Tests (CAT)	-	1
Mobility Questionnaire 47 (MobQues47)	-	1
Quality of Life		
Child Health Questionnaire (CHQ)	2	-
Pediatric Quality of Life Inventory (Peds- QoL)	1	-
Cerebral Palsy Quality of Life for Children (CP- QoL)	1	-
Multi-Dimensional Outcome Measures		
Patient-Reported Outcomes Measurement Information System (PROMIS)	-	1

Adverse event and harm		
Clavien-Dindo System	-	3

Table 69 Outcome measures used both before and after 2016

Outcome measures	**Pre- 2016** (6, 191) N = 310	**Post- 2016** (227) N = 44
	n (%)	**n (%)**
Body Function and Structure		
Clinical examination	208 (67)	31 (70)
Gait analysis (kinematics with or without kinetics)	192 (62)	37 (84)
Gait Velocity	95 (31)	8 (18)
Gait Profile Score (GPS)	11 (4)	8 (18)
Gait Deviation Index (GDI)	20 (6)	12 (27)
Gillette Gait Index (GGI)	20 (6)	1 (2)
Radiology	77 (25)	7 (16)
Foot pressure data	8 (3)	3 (7)
Oxygen consumption	6 (3)	2 (5)
Timed Up and Go (TUG)	1 (1)	1 (2)
Activity and Participation		
Gross Motor Function Measure (GMFM)	23 (7)	2 (5)
Pediatric Evaluation of Disability Inventory (PEDI)	1 (1)	1 (2)
Gillette Functional Assessment Questionnaire (Gillette FAQ)	13 (4)	3 (7)
Functional Mobility Scale (FMS)	16 (5)	1 (2)
Pediatric Orthopaedic Data Collection Instrument (PODCI)	10 (3)	3 (7)

11.3.3 Outcome measures screening

In total, 424 eligible studies were identified in three reviews, and 38 different outcome measures were identified. The output of the initial screening of the identified outcome measures against the pre-specified criteria was a list of outcome measures (n= 20) that could be considered as a set of potential core outcome measures. The list is shown in Table 70.

Table 70 Outcome measures used in clinical trials of lower limb orthopaedic surgery for children with CP. Gait analysis is represented by the three most commonly used gait indices (GGI, GDI and GPS).

Outcome measure	Wilson et al. (2014)(6)	Wilson et al. (2015)(191)	Almoajil et al. (2020)(227)	Inclusion	Exclusion/ justification
Body Function/ Structure					
Clinical examination	x	x	x		Part of Gait Analysis
Gait analysis					
GGI	x	x	x	In	
GDI	x	x	x	In	
GPS	x	x	x	In	
Gait velocity	x	x	x		Part of Gait Analysis
Movement Analysis Profile			x		Part of Gait Analysis and summarised kinematic data from GPS
Edinburgh Visual Gait Score			x	In	
Radiology	x	x	x		Any abnormal post-operative radiology would reflect on the recording of adverse events
Type of walking device	x				Subjective
Surface electromyography	x	x			Part of Gait Analysis
Presence of pain	x	x			Subjective – requires appropriately validated pain scale to be measured
Foot pressure data/ Foot progression angle	x	x	x		Part of Gait Analysis
Physiological Cost Index	x				Part of Gait Analysis
Energy cost of walking/ oxygen consumption	x				Part of Gait Analysis
Biomechanical Model	x				Part of Gait Analysis
Normalcy Index	x				Part of Gait Analysis and similar to GGI
Hip Flexor Index	x				Part of Gait Analysis
Observation Gait Score	x			In	
Vertical plantar pressure	x				Equivalent to Foot pressure data
Stahelil's Rotational Profile			x		Limited use
Selective Control Assessment of the Lower Extremity	x				
Total mechanical work		x			
Physician Rating Scale	x			In	
Timed Up and Go		x	x	In	
Activity and participation					
Gross Motor Function Measure	x	x	x	In	
Gillette Functional Assessment Questionnaire	x	x	x	In	
Functional Mobility Scale	x	x	x	In	
Functional Independence Measure for Children	x	x		In	
Positional Activity Logger	x				Subjective
Pediatric Evaluation of Disability Inventory	x	x	x	In	

Mobility Questionnaire 47			x	In	
Modified Goal Attainment scale	x				The purpose of the measurement instrument was to set a goal only for general CP population
Gross Motor Performance Measure	x			In	
Quality of life					
Child Health Questionnaire	x			In	
Pediatric Quality of Life Inventory	x			In	
Cerebral Palsy Quality of Life for Children		x		In	
Multi-dimensional measures					
Pediatric Outcomes Data Collection Instrument	x	x	x	In	
Computerised adaptive test			x		Part of PROMIS
PROMIS			x	In	
Further outcome measures (newly developed outcome measurer)					
GOAL				In	

There were 20 shortlisted outcome measures, each of which demonstrated evidence of reliability and validity. The majority of the measures were generic (n= 16) and clinician-driven measures (n= 11) and three were patient or proxy-reported only; PEDI, MobQues74 and the GOAL questionnaire. Most of the measures were applicable to children and young adults with CP aged 0-18 years; only three measures (GMFM, GMPM and CP-QoL) were specifically for children ≤ 12 years of age. Gait Analysis was assessed in terms of the most commonly used gait indices (GGI, GDI and GPS).

11.3.4 Mapping to the Core Outcome (Domains) Set

The outcome measures were linked to the domains and outcome levels of the COS for lower limb orthopaedic surgical interventions that were identified earlier (Chapter 10). The 20 measures varied considerably in the degree to which their content represented the contents of the COS. Table 71 illustrates the mapping of the potential measures to the core outcomes (domains) set.

Primarily, measures covered the mobility domain. Daily life activities, climbing stairs and walking-related outcomes were the areas most frequently represented by the measures. The evaluation measures that demonstrated the greatest coverage of the outcomes that had been included in the core outcome (domains) set were the GOAL and PODCI measurement tools.

Table 71 Outcome measures aligned with the core outcome (domains) set. Gait analysis is represented by the three most commonly used gait indices (GGI, GDI and GPS).

Domain	Outcomes	Gait Analysis			EVGS	PRS	OGS	FMS	GMFM	TUG	FAQ	PEDI	FIM	GMPM	MobQues	PODCI	PROMIS	GOAL	CHQ	CP-QoL	PedsQL
		GPS	GDI	GGI																	
Pain, discomfort, and fatigue	Pain															X	X	X	X	X	X
	Muscle fatigue																	X			
	Energy level after activities															X	X	X			X
Lower limb structure	Alignment and symmetry					X		X										X			
Motor function	Standing position																	X			
	Fall and balance								X	X								X			X
Mobility: Daily Life Activity	Basic life activities								X		X	X	X	X	X	X	X	X			
	Climbing stair								X		X		X		X	X	X	X			
Mobility: walking	Walking speed									X								X			
	Walking distance							X		X	X					X		X			
	Walking endurance											X			X	X		X			
	Walking pattern (quality)	X	X	X	X	X	X											X			
	Walking ability								X				X		X	X		X			
	Using orthosis, splint and brace															X		X			
	Using assistive device							X				X				X		X			
Participation	Physical activity										X					X	X	X			
Independence	Independence												X					X			
Quality of Life	Self-esteem																	X	X	X	
	Well-being																		X	X	X

11.3.5 Quality assessment

COSMIN Database

The COSMIN database search initially identified 58 reviews. The titles and abstracts of these reviews were extracted and screened. Of these, 49 were excluded: five used different appraisal tools (e.g., CanChild rating form), 12 did not provide data regarding relevant outcome measures, two were studies of primary psychometric properties, 20 were out of the current review scope, six were structured/ scoping reviews of literature, one was based on outcome measures in the adult CP population, and three reviews did not specify the health conditions that had been involved in the studies (e.g., children with neurological disorders). An example of the excluded reviews can be found in Appendix 14. This process left nine reviews for inclusion. Table 72 provides a summary of the characteristics of the included reviews.

The identified reviews were published between 2008 and 2020. The majority of the reviews were related to children with CP (n= 5). The reviews had diverse focus: three targeted gait- and walking-related outcomes, two aimed to assess quality of life outcome measures, two reviews focused on outcome measures of motor function, one focused on balance and one focused on daily life activities.

Table 72 Reviews identified from COSMIN database

Author/ year	Title	Condition	Review Focus	Outcome measures	Ref
Pavao et al (2017)	Clinical tools designed to assess motor abilities in children with cerebral palsy	CP	Motor abilities	GMPM, PEDI, FIM	(535)
Saether et al (2013)	Clinical tools to assess balance in children and adults with cerebral palsy: a systematic review	CP	Balance	TUG	(536)
Ferre-Fernándes et al (2020)	Measures of Motor and Functional Skills for Children with Cerebral Palsy: A Systematic Review	CP	Motor function and skills	GMPM, FIM	(537)
Himuro et al (2017)	Easy-to-use clinical measures of walking ability in children and adolescents with cerebral palsy: a systematic review	CP	Walking ability	TUG, FMS, Gillette FAQ	(527)
Zanudin et al (2017)	Psychometric properties of measures of gait quality and walking performance in young people with Cerebral Palsy: A systematic review	CP	Gait quality and walking performance	3DGA (GDI, GPS), OGS, EVGS, PRS, TUG, FMS, Gillette FAQ	(219)
Debuse et al (2011)	Outcome Measures of Activity for Children with Cerebral Palsy: A Systematic Review	CP	Activity	GMFM, PEDI	(538)
Ammann-Reiffer et al (2014)	Measurement properties of gait-related outcomes in youth with neuromuscular diagnoses: a systematic review	Neuromuscular diagnoses	Gait function	MobQues47	(539)
Janssens et al (2015)	A Systematic Review of Generic Multidimensional Patient-Reported Outcome Measures for Children, Part II: Evaluation of Psychometric Performance of English-Language Versions in a General Population	Children and young adult (all health condition)	Quality of Life	CHQ, PedsQL	(289)
Coombes et al (2016)	Health-related quality-of-life outcome measures in paediatric palliative care: A systematic review of psychometric properties and feasibility of use	Paediatrics	Quality of Life	CP-QoL	(540)

PubMed Database

The PubMed search was conducted for reviews of the psychometric properties of three outcome measures: GOAL, PODCI and PROMIS. The literature search initially identified 13 studies. A manual search of the references that were cited in the available reviews of CP yielded an additional eight abstracts. All identified studies were imported into an EndNote library (Endnote X8), duplicates were removed (zero) and the remaining 21 titles/abstracts were screened; of these, 16 were selected for full-text review. Four of these were excluded: one was a commentary, the data for one study were unavailable, and two studies were prospective and therefore they did not report any psychometric property data. Therefore, 12 studies of reported psychometric properties were considered.

Table 73 provides a summary of the characteristics of the included studies. The distribution of the studies includes four studies related to the GOAL questionnaire, five studies reported data for the PODCI, and three studies reported data for the PROMIS. The majority of the studies involved participants between 2 and 25 years of age. In terms of sample size, all studies involved more than 30 participants. Reliability was assessed five times for all outcome measures; validity was assessed 14 times; and three studies aimed to assess the responsiveness of each measure.

The methodological quality of the studies that had assessed the psychometric properties of the three outcome measures was rated as 'excellent' (15 times), 'good' (5 times) and 'poor' (2 times). The reasons for the 'poor' rating, referring to the COSMIN checklist, were insufficient information regarding the study design in one study and the use of inappropriate statistical analysis in the other study. In terms of adapted quality criteria [18], most of the psychometric properties of the outcome measures in this review were rated as 'positive' (18 times), with the remainder being rated as intermediate (1 time) and 'negative' (3 times). The inter-rater reliability of the PODCI to determine functional health post-surgery was scored as 'negative' since the intraclass correlation coefficient (ICC) was below <0.70. Similarly, 75% of the PROMIS data were not in accordance with the hypothesis that was under testing, and in line with good measurement criteria, a 'negative' score was applied. Finally, the structure validity of PROMIS was rated 'negative' because the authors found DIF by different levels of mobility.

Table 73 Characteristics of the included studies and ratings of methodological quality and quality of evidence

Outcome measure	Study	n, age (y)	Psychometric properties	Results	(COSMIN) rating	Quality of Evidence	Reference
GOAL	Thomason et al (2018)	105, 6-20	Hypothesis testing: discrimination validity	(Mean [SD]) GMFCS level I, 72.5 [12.7]; GMFCS level II, 61.4 [13.0]; GMFCS level III, 38.8 [10.6]; [F2,97=42.4, p<0.001]) FMS (5m and 50m r=0.59; 500m r=0.66)	Excellent	+	(3)
			Construct validity	FAQ walking (r=0.77) and activities list (r=0.75, p<0.01). GPS (r=0.59) and gait appearance domain and GPS (r=0.52, p<0.01)	Excellent	+	
	Thomason et al (2020)	43, 6-23	Responsiveness	Surgical group: mean (SD) 61.7 (18.1) to 63.8 (16.1) Non-surgical group: mean (SD) 65.5 (15.6) to 64.6 (16.4) Correlation between change in total GOAL and GPS for all children was −0.42. The strongest correlation was −0.47 between the gait appearance domain and GPS	Good	+	(541)
	Karpati et al (?)	51, 7-16	Content validity	Children's priorities correlated with their parents'; however, parents rated all items as more important goals than their children	Poor	+	(542)
	Boyer et al (2021)	310, 3-25	Hypothesis testing: convergent validity	95% CI GMFCS I-IV= 0.73-0.84	Excellent	+	(543)
			Construct validity	95% CI GMFCS I-IV= 0.24-0.49	Excellent	+	
PODCI	Damino et al (2005)	35, 4-16	Responsiveness	Surgical group: the GMFM Total score was responsive to change after both MT surgery, p < 0.05 for pre/postsurgery. Significant correlations found between PODCI Global Function Scale and Subscales and GMFM Scores were mostly in the moderate to high range (0.64-0.94), with the highest value seen between the GMFM Total Score and the Transfers and Basic Mobility Subscale.	Excellent	+	(544)
	Deltroy et al (1998)	470, 2-8	Internal consistency	Cronbach's alpha 0.76–0.97	Excellent	+	(545)
			Interrater reliability	0.45–0.87, highest for physical domain (parent vs child)	Good	-	
			Test-retest reliability	0.71–0.9	Good	+	
	McCarthy et al (2002)	115, 3-10	Hypothesis testing: convergent validity	Spearman's partial correlation coefficients 0.60–081 (CHQ), 0.56–0.94 (GMFM), 0.50–0.81 (PEDI)	Excellent	+	(546)

			Internal consistency	Cronbach's alpha 0.76–0.97	Excellent	+	
			Hypothesis testing: discrimination validity	Detect change according to severity: The transfer and mobility subscale (RV, 62%)	Excellent	+	
	Pencharz et al (2001)	166, 5-17	Construct validity: convergent validity	Spearman's partial correlation coefficients CHQ: r ≥ 0.68 (pain subscale) CHQ: 0.21 ≤ r ≤ 0.56 (global function) ASK: r ≥ 0.78	Good	+	(547)
			Hypothesis testing: discriminative validity	Detect change according to topographic: F test (74.8) 0.0001 ≤ p ≤ 0.005	Excellent	+	
	Vitale et al (2005)	180, 5-18	Hypothesis testing: convergent validity	Sensitive to differences in the diplegic and hemiplegic groups	Excellent	+	(548)
			Criterion validity	% Agreement (physical score): 33% - 41% (except diplegia) % Agreement (psychological score): 8% - 35%	Poor	?	
PROMIS	Kratz et al (2013)	82, 8-19	Internal consistency	Cronbach's alpha 0.75	Excellent	+	(549)
			Construct validity: convergent validity	Scores of PROMIS (short form) and PROMIS mobility CAT, respectively PedsQL: 0.58-0.60 PODCI (mobility): 0.52-0.39 PODCI (sport): 0.60-0.49 Gillette FAQ: 0.48-0.41 GMFM (stand): -39-0.21) GMFM (walk): 0.39-0.19 TUG: -30 - -0.16	Excellent	+	
			Hypothesis testing	PROMIS (Short Form) and all legacy measures were able to distinguish between GMFCS categories F (2,81) =9.55 p <0.001 PROMIS (CAT) scores did not show significant mean differences between the groups F (2,78) = 2.90 p=0.06	Excellent	-	
	Coster et al (2016)	303, 8-17	Structure validity	Fatigue, M2=1050.25, (RMSEA)=0.04; Pain Interference, M2=806.99, RMSEA=0.05. Mobility, M2=638.6, RMSEA=0.04.	Good	-	(550)
	Mulcahey et al (2016)	Baseline: 113, 12.7 (3.0) 6-month: 98, 12.7 (3.1) 12-month: 86, 12.4 (3.0)	Responsiveness	The PROMIS CAT and SF did not detect change at either the 6- or 12- months. ES and SRM for all PROMIS Pediatric Measures were non-significant.	Excellent	+	(551)

Best Evidence Synthesis

Table 74 shows the 20 outcome measures for psychometric criteria based on the best evidence synthesis. With the exception of reliability, internal consistency, hypothesis testing and responsiveness, the majority of evidence for measurement error, structure, criteria and cross-cultural validity is unknown. PEDI demonstrated the greatest amount of positive psychometric evidence across the seven areas of reliability, validity and responsiveness domains. Twelve outcome measures showed strong to moderate positive evidence in one or more measurement properties: the GPS, GMFM, FMS, Gillette FAQ, PEDI, FIM, MobQues47, TUG, PedsQL, GOAL, PODCI and GMPM. Five outcome measures had conflicting evidence to support positive results for reliability: GGI, PRS, CHQ, PedsQL and PODCI. There were two negative pieces of evidence across all measurement properties of all outcome measures. These were the criterion validity of the Observational Gait Scale (OGS) and measurement error of the CHQ. Evidence for responsiveness of any outcome measures in this population was either poor or unknown, with the exception of the GMFM, FMS, PEDI, GOAL, PODCI and PROMIS.

11.3.6 Feasibility (clinical utility)

Feasibility criteria were extracted for the 20 outcome measures (Table 75). Nine measures (45%) had been administered via proxy-report, and six of them (30%) included a self-report version through which children and young adults could share their perspectives. Eleven measures (55%) were based on administration by a clinician.

In terms of complexity and the time taken for administration of the measures, the GMFM, GMPM, PEDI and FIM scales required at least 30 minutes for administration and were more complex than the others in terms of set up and/or scoring. However, these measures provided more comprehensive information regarding the child's function. On the other hand, simple measures required 20 minutes or less for administration. These were: the Gillette FAQ, FMS, MobQues47, TUG, GOAL, PODCI, CHQ and CP-QoL.

Table 74 Psychometric properties of the shortlisted outcome measures. Gait analysis is represented by the three most commonly used gait indices (GGI, GDI and GPS).

Outcome Measure		Reliability			Validity					Responsiveness	References
		Reliability	Measurement error	Internal Consistency	Content validity	Structure validity	Hypothesis testing	Cross-cultural validity	Criterion validity		
Gait Analysis	GGI	±	?	0	0	0	0	0	0	0	(219)
	GDI	?	?	0	0	0	+	0	0	?	(219)
	GPS	0	0	0	0	0	++	0	0	0	(219)
EVGS		?	0	0	0	0	+	0	?	?	(219)
OGS		?	0	0	0	0	0	0	-	0	(219)
PRS		±	0	0	0	0	0	0	0	0	(219)
GMFM		+++	0	0	++	0	+	0	0	+++	(538)
FMS		++	0	0	0	0	?	0	?	+	(219, 527)
Gillette FAQ		++	0	0	0	0	++	0	?	0	(219, 527)
PEDI		+++	0	+++	++	+	+++	+	0	+++	(535, 538)
GMPM		++	0	0	0	0	+++	0	0	?	(535, 537)
FIM		+	0	+++	0	+++	+	0	0	0	(535, 537)
MobQues47		++	?	0	0	0	0	0	0	0	(539)
TUG		++	?	0	+++	0	++	0	?	?	(219, 527, 536)
CHQ		±	-	+	0	±	±	0	0	0	(289)
PedsQL		±	±	+	0	++	±	0	0	?	(289)
CPQoL		+	0	?	0	?	+	0	0	0	(540)
GOAL		+	0	+	?	0	+++	0	0	±	*
PROMIS		0	0	+	0	0	+	+	0	+	*
PODCI		±	0	+++	0	0	+++	0	?	±	*

* There are no existing COSMIN reviews available, and researchers have assessed the quality of the evidence.

Table 75 Feasibility criteria of the shortlisted outcome measures. Gait analysis is represented by the three most commonly used gait indices (GGI, GDI and GPS).

Outcome Measure		Administration format	Age	Administration time	Length of survey	Equipment needed	Assessor training	Cost	References
Gait Analysis	GGI	Clinician	Not specified	Not specified	16 items	Gait lab, software	Yes	Lab: Yes	(552)
	GDI	Clinician	Not specified	Not specified	9 items	Gait lab, software	Yes	Lab: Yes	(552)
	GPS	Clinician	Not specified	Not specified	9 items	Gait lab, software	Yes	Lab: Yes	(552)
EVGS		Clinician	Not specified	Not specified	17 items	Video recordings	Yes	Software: Yes	(175, 553, 554)
OGS		Clinician	Not specified	Not specified	Not specified	Video recordings	Yes	Free	(175, 553)
PRS		Clinician	Not specified	Not specified	6 items	Video recordings	Yes	Free	(175, 553)
GMFM		Clinician	0-12 y	45-60 min	66 items	Computer, software	No	Manual: Yes	(141, 555)
FMS		Clinician	4-18 y	5-10 min	3 items	No	No	Free	(184, 527)
Gillette FAQ		Self- Proxy-reported	Not specified	5 min	22 items	No	No	Free	8, 24
PEDI		Proxy-reported	6m-8 y	30-60 min	197 items	Software	No	Yes	(141, 556)
GMPM		Clinician	5m-12 y	45-60 min	20 items	Computer, software	Yes	Yes	(535)
FIM		Clinician	7-18 y	30-45 min	18 items	No	No	Free	(141, 190)
MobQues47		Proxy-reported	2-18 y	15-20 min	47 items	No	No	Free	(557)
TUG		Clinician	3-18 y	5 min	5 items	Standard armchair, stopwatch	No	Free	(527)
CHQ		Self- Proxy-reported	Self-report: 10-18 y Proxy-report: 5-18 y	Self-report: 16-25 min Proxy-report: 5-15 min	Self-report: 87 items Proxy-report: 28-50 items	Computer	Minimum training on scoring	Free	(558, 559)
PedsQL		Self- Proxy-reported	Self-report: 5-18 y Proxy-report: 2-18 y	5-10 min	23 items	None	No	Yes	(141, 287, 560, 561)
CP-QoL		Self- Proxy-reported	Self-report: 9-12 y Proxy-report: 4-12 y	15-25 min	Self-report = 53 items Proxy-report = 66 items	No	No	Free	(198, 562)
GOAL		Proxy-reported	9-18 y	12-19 min	48 items	Form	No	Free	(3, 563)
PROMIS		Self- Proxy-reported	Self-report: 8-17 y Proxy-report: 5-17 y	Not specified	Not specified	Computer	No	Yes	(141, 530, 564, 565)
PODCI		Self- Proxy-reported	Self-report: 8-17 y Proxy-report: 2-18 y	10-18 min	Self-report: 83 items (11–18 y) Proxy-report: 86 items (11-18 y) Proxy report: 86 items (2-10 y)	Computer	No	Free	(141, 564)

11.3.7 Consensus meeting

The consensus group recommended that the outcome measures shown in Table 76 could be used as optimal options, where possible, in studies of lower limb orthopaedic surgery for ambulant children with CP. Because there were high numbers of outcomes within domains, it was agreed that the production of a single measure for each outcome domain would not be possible. After the whole group discussion, it was agreed that the GOAL measure was a comprehensive tool that captured all of the core outcome domains and can be recommended and used as the optimal available option (compared to other existing measures) for use in future studies. Other outcome measures were also discussed (Appendix 15).

For the 'pain, discomfort and fatigue' core outcome, two measures reached consensus: the GOAL and the PROMIS. The GOAL questionnaire was favoured over the use of the PROMIS since the GOAL was a condition-based measure and, importantly, it could be used as an instrument to evaluate the effectiveness of surgical interventions. Although condition-specific measures might be more sensitive to change, the PROMIS, which is a generic measure, was suggested as a preferred measure when comparison of ambulant CP against different paediatric orthopaedic conditions was necessary. The consensus group felt that the construct of precise pain location and pain interference with the child's daily life were essential aspects of pain, as these elements would help with clinical decision-making on treatment options. These aspects were lacking in the GOAL and the PROMIS.

The 'lower limb structure' domain describes the appropriate alignment and symmetry of the lower limbs, both functionally and cosmetically. A consensus was reached that the GOAL could be recommended and used as the most optimal of available options. The GOAL covers aspects of upright and crouched postures, and foot positions during activities. For the recommended core outcome domain of 'motor function', consensus was reached early that the GMFM-66 was preferred, followed by the GOAL as a proxy-reported measure. Panel members reflected on the time that was necessary to complete assessments with the GMFM-66 and the training required to use it. However, the panel felt that the reliability, validity and depth of the GMFM-66 content to capture outcomes in the motor function domain outweighed these disadvantages.

The 3DGA, the Gillette FAQ and the GOAL questionnaire achieved the panel consensus as clinician-driven and PROMS that were important for the measurement of the 'mobility' domain. Mobility covers aspects of daily life activities and gait-related outcomes (e.g., walking speed, distance and pattern). The 3DGA is

considered the gold standard in surgical decision-making for CP since it provides objective gait data, including comprehensive kinetic and kinematic parameters. However, as the measure requires use of an expensive and complex laboratory, the panel agreed that the EVGS was a useful alternative measure. It should be noted that the output of valid and reliable 3DGA data depends on optimal quality control of the laboratory that produces them. Therefore, this should be taken into account when considering 3DGA as an outcome measure. Furthermore, the specific usefulness and role of each individual gait parameter or gait index as a potential outcome has not been fully explored.

The Gillette FAQ and the GOAL both capture important aspects of mobility outcomes. The panel members recommended the GOAL measure over the Gillette FAQ since the former provided more items that investigated the level of assistance that was required for different activities and covered the orthotics aspects, which was an outcome that was identified as important by the children with CP. Some panel members suggested a new outcome measure; 10-metre walk test (10-MWT) as a superior tool to measure walking speed compared with the TUG test. They argued that both tests measured capacity level, the TUG tested basic balance and mobility skills rather than walking speed in isolation. Although the 10-MWT was preferred, the research team had not explicitly considered the psychometric properties during measurement appraisal, and they found that the level of evidence remained unclear at the time of the meeting. Therefore, the quality of the psychometric properties and feasibility of the use of the 10-MWT were later considered by the research team and it was subsequently included in the COS.

The panel chose to recommend measurement of ‘physical activity’ and ‘independence’ through the recently developed patient and proxy-reported outcome measure, the GOAL. Although no study had specifically identified measurement of physical activity or independence associated with lower limb orthopaedic surgery, the GOAL had shown good internal consistency, inter-rater reliability and validity. The questionnaire consists of eight items that track physical activities and 19 items that assess the level of assistance that is required in daily life activities and gait-related outcomes, which can be computed as independence.

The panel explored the difficulties of measuring quality of life due to its multi-dimensionality, although they felt that self-esteem and body image contributed to it. The group discussion focused on the variability of domains within quality of life outcome measures and agreed that no single outcome measure could cover all aspects of quality of life. They emphasised that different outcome measures could be suggested and used in different settings according to the research question under study. Since

the panel felt unable to state a preferred outcome measure specific to quality of life, two outcome measures reached consensus approval: the GOAL and the CP-QoL questionnaires. The CP-QoL list is a comprehensive tool that tracks quality of life in relation to general health, participation, communication, pain, family and friends, school, use of special equipment, and access to services. The group consensus was to use the PROMIS to measure quality of life via the social health domain, which was considered to impact on quality of life. The PROMIS was recommended for use in studies that aim to compare quality of life across paediatric conditions, as it was considered a generic measure.

Measurement of adverse events was considered separately. Although no specific outcome measures that had been designed specifically to assess surgical adverse events (i.e., complications) had been identified during the previous steps, use of the Clavien-Dindo System was highly recommended to grade surgical adverse events in future study reports and documentation.

Table 76 Core outcome measurement set

Outcome Domain	Outcome measure*
Pain, discomfort and fatigue	- Gait Outcomes Assessment List (GOAL) - Patient-Reported Outcomes Instruments System (PROMIS)
Lower limb structure	- Gait Outcomes Assessment List (GOAL)
Motor function	- Gross Motor Functional Measure (GMFM-66) - Gait Outcomes Assessment List (GOAL)
Mobility: Daily life activities	- Gait Outcomes Assessment List (GOAL)
Mobility: Gait-related outcomes	- Three-Dimensional Gait Analysis (3-DGA) - Edinburgh Visual Gait Score (EVGS) - Gait Outcomes Assessment List (GOAL) - Gillette Functional Assessment Questionnaire (Gillette FAQ) - 10-Meter Walk Test (10-MWT)
Physical activity	- Gait Outcomes Assessment List (GOAL)
Independent	- Gait Outcomes Assessment List (GOAL)
Quality of Life	- Cerebral Palsy- Quality of Life (CP-QoL) - Gait Outcomes Assessment List (GOAL) - Patient-Reported Outcomes Instruments System (PROMIS)

* Refer to tables 74 and 75 for outcome measures characteristics, properties, and references.

11.4 Discussion

The aim of the study was to identify and select the most optimal among existing outcome measures for use to assess lower limb orthopaedic surgical interventions for ambulant children with CP in research. This study moves towards the standardisation of outcome measures in this field.

Identified outcome measures

This updated review considered additional studies to those that had been considered in the first review by Wilson et al., which was conducted in 2016. The additional studies contained clinical information on the outcome measures that were used to assess the effect of lower limb orthopaedic surgery in CP. The revised search yielded 29 outcome measures, of which nine were unique to this review. These findings can be attributed to the continued development and expansion of CP outcome measures.

The findings of the scoping review identified some trends in the use of PROMs for lower limb surgery, that are available in this field, covering the majority of the outcome domains including pain, mobility, walking ability, independence and social function. The PODCI and Gillette FAQ were the most commonly used PROMs in the studies that were identified in this review. This was probably because PODCI could be used to assess the impact of surgery across multiple domains of functioning (physical, mobility, and sport), as well as pain and happiness. The Gillette FAQ is used to assesses walking ability and mobility domains, which are recognised as important domains for children and their parents (7, 225).

PROMs do not assess certain key outcomes such as joint RoM, spasticity and gait pathology, which are usually assessed by the clinicians, as discussed in section Chapter 3. Therefore, PROMs could be viewed as complementary measures to be used together with clinician-reported measures. Although several PROMs provide a self-assessment from the perspectives of children or proxies, they differ in their levels of objectivity, the outcome that is assessed, their feasibility and cost of administration. Little standardisation was found across the five studies in the use of PROMs, which limited the researcher's ability to synthesise studies to explore the effectiveness of surgical interventions (10).

The review identified limited reports of adverse events. This sparsity of reporting may have indicated selection bias in reported outcomes within studies (5, 110). Repeated surgeries and side effects (e.g., infection, neurovascular complications) were reported as adverse events of surgery; however, these adverse outcomes accounted for less than 25% of the total outcomes that were identified, and they were reported in only 15 of 44 studies (34%). This imbalanced reporting between the benefits and harms can lead to a poor understanding of the impact of surgery on an individual's health (423). Furthermore, clear knowledge of postoperative complication risk is important since it can both inform surgery choice by clinicians and is considered a key element that the patient must understand before giving consent for surgery (424).

The heterogeneity in outcome measures (n= 29) across studies was probably compounded by the variation in follow-up periods across studies. The maintenance of the surgical outcomes at different time points should be investigated, but, consistency in follow-up time points across studies is recommended to support comparisons in future systematic reviews and meta-analyses (566). Future studies are needed in order to identify the most clinically meaningful follow-up time points, since the full effects of surgery on a child's level of activity and function may not be apparent for a long time.

Recommended outcome measures

The outcome measures recommended by the consensus group were a combination of clinician-driven measures (objective): the 3-DGA, EVGS, 10-MWT and GMFM; and of PROMs: the GOAL, Gillette FAQ, PROMIS and CP-QoL.

Schiariti et al. (141) and Wright et al. (567) suggested that such a combination represents appropriate pairing of outcome measures, and in particular, they highlighted the growing importance of the inclusion of PROMs in the assessment of CP. Some measures, when used alone, may fail to capture the real impact of surgery on the child due to the determinants that may influence the outcomes of interest. One example is the measurement of gait deviation through the use of 3DGA, which is a major component of gait-specific outcome measurement and is important in surgical decision-making. Studies show that changes in magnitude of the deviation from normal gait through 3DGA are correlated moderately or poorly with patient satisfaction and their perceived quality of life during the first year after the operation (123). Therefore, to assess the multivariable aspects of the surgical intervention and to reflect performance in a real-life context, PROMs could occupy a central place in the selection of outcome measures alongside clinician-driven measures when future clinical trials are designed.

The preferred and recommended outcome measure within the consensus panel was the GOAL questionnaire. The Gait Outcomes Assessment List (GOAL) questionnaire was developed as a comprehensive assessment tool according to the ICF-CF framework for ambulant children with CP. It was developed by a multidisciplinary team in Toronto, Canada, with direct input from children with CP and their parents. It measures gait priorities and other functional mobility outcomes. There are two versions of the questionnaire for parent and child. The GOAL questionnaire comprises 49 items grouped into seven domains: (1) Activities of daily living and independence (9 items), (2) Gait function and mobility (10 items), (3) pain, discomfort and fatigue (7 items), (4) physical activities, sports and recreation (8

items), (5) gait pattern and appearance (6 items), (6) use of brace and mobility aids (3 items) and (7) body image and self-esteem (6 items).

The recommendation of using multiple outcome measurements was welcomed, as a development that better-reflected outcomes across the ICF-CY framework, recommended in previous reviews in this field. For example, the review by Wilson et al. (6) of outcome measures stated: "We suggest that in order to understand the full impact of lower limb orthopaedic surgery, a suite of outcome measures across the ICF may be needed, including the domain of activity and participation that reflects outcomes relevant to the patient, family, and surgeon". Watkins et al. (568) stated that "no single instrument can be used to measure all of the components of the ICF" when they reviewed functional and health-related quality of life measures for children with CP, and recommended the use of multiple measures to cover aspects of the ICF.

The reliability and validity of the GOAL have been rated several times as 'limited' or 'unknown'; however, this is to be expected since it was a newly developed measure at the time of this review, and there was a lack of primary evaluation of its psychometric properties. The acquisition of high COSMIN rating relies on the accumulation of high-quality evidence. Despite the level of evidence, the inclusion of the GOAL measure (3), which assesses multidimensional domains, is viewed for two reasons as a strength of the recommendations that were made from the core group of outcome measures. First, the use of the measure enables the research/practice agenda to move beyond a focus on symptom management/disability and to consider the role of the surgery in other health domains such as gait-related outcomes, physical activity and the quality of life of children with CP. Second, it reflects the awareness of researchers and clinicians regarding the outcomes that are relevant to the patient and family, that is, towards activity and participation.

It is also of note that, with the exception of the GOAL, all the core outcome measures that were recommended for measurement of quality of life are contained in the review by Watkins and colleagues of recommended outcome measures (of which there are four recommended quality of life measures) (568). The omission of the GOAL from the Watkins et al. recommended measures could be a reflection of the fact that it had been recently developed at the time of the recommendation. Furthermore, the proposed core outcome measurements set includes several of the multiple-item outcome measures that were suggested by Schiariti and colleagues for children with CP, in particular the GMFM, Gillette FAQ and CP-QoL (285).

Data on the psychometric properties of several outcome measures had not been evaluated. This is an important consideration in clinical care. For example, 45% of the identified measures had been used as evaluative measures although they had not been tested for responsiveness to change (e.g., MobQues47, FIM). Health professionals need to be aware of these important psychometric properties to avoid misuse of measurements and misinterpretation of results. Further studies can build upon these identified gaps in the literature to investigate other important psychometric properties.

Apart from 3DGA, the core outcome measures that were recommended are commonly used assessments that can be completed relatively quickly, require no specialised equipment or training and have minimal cost implications. It is feasible to use these outcome measures in future studies of lower limb orthopaedic surgery. It is not intended that these recommended measures should be the only ones for use in future studies, but that these measures may be supplemented with other measures according to the purpose of the study, e.g., if the study is for exploratory purposes or to evaluate condition-specific issues.

In light of the evolving evidence base, the publication of new outcome measures and the continued validation/update of existing outcome measures, the proposed core set of measures must be reviewed regularly. During such review, the results of recent studies may provide stronger evidence to support the inclusion of these proposed measures or additional/different ones. Furthermore, while kinematic and kinetic parameters and their derivatives in the form of GPS and GDI values are the most commonly used gait measures, more research is needed to determine the most relevant gait parameters derived from the 3DGA, and the clinical examination as outcome measures. It should also be born in mind that the validity and reliability of 3DGA measurements depends on the quality control practices of the relevant labs producing them. Critical appraisal and scientific rigor in evaluating these parameters is, therefore required.

Strengths and limitations

A strength of the study was that that the consensus recommendations were made based on best evidence that included recently published systematic reviews on the measurement properties and feasibility criteria. Moreover, the consensus methodology was informed by existing recommendations and guidelines, such as those produced by the COMET and COSMIN initiatives.

There were four main limitations to our study. First, the consensus meeting panel was small and its members were predominantly from the UK and English-speaking countries. This may have impacted the consensus process and findings. Second, patients were not involved in the consensus procedure, as this is not part of the COMET/COSMIN recommended methodology. Patients and representatives (e.g., parents) could have provided useful comments on the friendliness and comprehension of the outcome measures. However, it should be stressed that it is unclear how patients can contribute to the selection of the core measurements set and take into account aspects such as measurement properties, and methodological research in this field is lacking. Third, all the core measures are available in English formats, and some have not been validated in other languages, which may limit the applicability of the core set and, particularly the PROMs. Research is necessary with respect to cultural adaptation and validation in a range of different languages to support the use of the core set in countries where English is not the primary language.

Fourth, performance of a half-day, online consensus meeting illustrated a challenge in the decision-making process to inform the choice of the core set. The panellists were experienced researchers and clinicians who had a good understanding of outcome measures and the science behind measurement of health. Yet the consensus was based on assumptions that the panellists had reviewed the quality of the measures prior to the meeting, since much discussion centred on the notion that the most suitable measures were those that covered the greatest number of ICF domains. It is likely that decision-making would have been enhanced by inclusion of face-to-face meetings with panellists, at which examination and discussion of the scientific quality of the measurements would have been the primary focus.

11.5 Conclusion

From the initial identification of existing outcome measures, it was evident that outcome domains were measured in different ways, with few consistently applied outcome measures. This heterogeneity confirmed the challenges in conducting a review of effectiveness and strengthened the justification for this work.

On the basis of this work, we would recommend that researchers should consider using a combination of PROMs and clinician-reported outcome measures following lower limb orthopaedic surgery to be conducted on ambulant children with CP. They should also consider practical aspects (e.g., costs) and availability of these measures in a particular language or country. The use of standard outcome measurements has the potential to enhance greatly the validity, comparability and clinical applicability of clinical trials on lower limb orthopaedic surgical interventions. Ongoing efforts are required to ensure global uptake of an agreed optimal core outcome measurements set.

Chapter 12 Discussion and Conclusion

The work described in this thesis was aimed to develop a COS for future effectiveness studies in lower limb orthopaedic surgery for children with ambulant CP. The methodology used and the findings of each stage of the study have been discussed in detail in preceding chapters. This chapter provides the context and rationale for this research and a summary of the key findings. It also highlights the contribution of this work to future research and the implications of its findings. Finally, it discusses the overall strengths and limitations of the identified COS, it suggests a dissemination plan and future research directions to build on this work.

12.1 Context of and rationale for this research

This research addressed the heterogeneity in reported outcome domains and outcome measurements in studies on lower limb orthopaedic surgery for ambulant children with CP. The wide variation of outcomes and outcome measurements used in previous research was a key factor that prevented systematic reviewers comparing the effects of specific surgical interventions for lower limb musculoskeletal problems (5, 110, 153, 216). The results of surgical studies in this field were often irrelevant to children or parents and the lack of patient-relevant outcomes affected the decision-making process (153, 209, 211).

The use of a COS is currently considered as the gold-standard method to overcome this limitation. COSs are increasingly recognised by key regulatory and research funding organisations, such as the NIHR, the HRA and NICE, as a key method to reduce heterogeneity in outcomes and to improve research quality (9, 215). Reducing the heterogeneity of reported outcomes and outcome measures should improve research quality in this field.

At the start of this project, there were key knowledge gaps in two areas. The first was the poor understanding of which outcomes all key stakeholders considered important in defining the overall success of lower limb orthopaedic surgery for ambulant children with CP. The second knowledge gap related to the tools used and the timing of measuring important outcomes. These knowledge gaps were addressed through the development of a COS recommended to be used in research studies on lower limb orthopaedic surgery in CP in order to improve the quality and relevance of evidence.

12.2 Summary of findings

The core outcome domains, and core outcome measurement sets are summarised here. There were five stages involved in the development of the core outcome domains set. The first three stages underpinned the exploration and definition of outcomes and outcome domains, while stages four and five described the steps taken to achieve consensus on which outcomes/domains should be part of a COS.

Development of the core outcome measurements set required four further stages. In stage one and two, the scope and exploration of outcome measures used in the literature were investigated. Stage three involved critical evaluation of the outcome measures that had been identified, based on psychometric properties and feasibility criteria. Finally, in stage four, consensus was achieved regarding which outcome measures would be recommended as part of the core outcome measurement set.

12.2.1 Core Outcome Domains Set

The first five stages of the study presented in Chapters 6-10 achieved the first aim of the project, the development of the COS domains for lower limb orthopaedic surgical intervention for ambulant children with CP. The five-stages are summarised below.

Exploration and definition of outcomes/domains

Qualitative Evidence synthesis: In Chapter 6, the qualitative evidence synthesis was described and addressed the objectives 1-A and 1-B. Only six qualitative studies transparently reported experiences and outcome expectations from individuals with CP and their families. The outcomes that were identified supported the assessment of activity, and participation and quality of life domains. No study was identified that had explored the views of health professionals on surgical outcomes.

Scoping review: In Chapter 7, the scoping review was described and aimed to support identification of a comprehensive list of the outcomes previously reported in the literature (objective 1-B). This review highlighted the significant heterogeneity in outcome reporting, which was evidenced by the 40 different outcomes that had been measured across 44 studies published within a three-year period. All studies reported outcome domains that considered body function and structure (100%), while only a few considered activity and participation (16%) and adverse events (30%). No single outcome was reported across all studies. Across the 44 studies, none included the assessment of health-related quality of life or independence.

Qualitative study: In Chapter 8, semi-structured interviews with 10 children and young adults with CP, eight families and 10 health professionals were described. This study aimed to identify what really mattered to stakeholders in terms of important outcomes of surgery and to further understand their experiences. This chapter addressed the project objectives 1-B and 1-C. Data were analysed through content analysis that was supported by ICF-CY linking rules and thematic analysis. Thirty-one outcomes were identified, which could be linked to 11 second-level ICF-CY categories: lower limb alignment and symmetry, flexibility and muscle strength, mental health, fatigue, pain, function in life, mobility, participation, being independent, quality of life, and adverse events caused by the surgery.

Compared to previous published studies, 10 new outcomes were identified by this study. This would suggest that some of the outcomes that are important to children and families have not been previously assessed. There is a great need to incorporate into future research the outcome domains that are important to all stakeholders, and the agreement of a COS would assist with this endeavour.

The qualitative study provided additional valuable insight into the factors that may facilitate or hinder achievement of these outcomes of importance. It led to the collection of a comprehensive list of 10 factors that included interdisciplinary collaboration, communication and information resources, holistic care and shared goal-setting, which were reported in this study as environmental facilitators of outcome expectations. In contrast, reported barriers included lack of time and resources to ensure thorough completion of rehabilitation e.g., insufficient physiotherapy sessions, and divergent expectations of professionals, children and families. Personal motivators included family encouragement, the patient's level of self-determination and previous experiences. Personal demotivators included fear of a new environment. These factors should further assist clinical reasoning and planning for surgical interventions for ambulant children with CP.

Approaching consensus methods: The combined "long list" of outcomes was discussed with the study team and steering committee to reduce the list of outcomes and to ensure clarity of the outcomes that were shortlisted for the Delphi process. A list of 37 outcomes were agreed to be included in the Delphi survey.

Achievement of consensus on the core outcome set

Delphi process: In Chapter 9, the international two-round Delphi survey was described. The survey was conducted to explore the most important outcomes for inclusion in the COS (objective 1-D). Participants were invited to rate the importance of 37 outcomes through the use of a nine-point GRADE scale. As discussed above, the list of outcomes was informed by the literature reviews and interviews with the key stakeholders and had been finalised and agreed by the steering committee. In the second-round survey, participants were invited to rank outcomes that had not reached consensus in the first round and four additional outcomes that were added in response to comments fed back by participants in the first round. After the two rounds of survey, 16 outcomes reached high levels of consensus across the stakeholder groups. Notable discrepancies were seen in opinions of eight outcomes, which reached consensus in the healthcare professionals' group only, and a further four outcomes that reached consensus in the individuals with CP and family group only. Conversely, four outcomes reached a consensus of limited importance in the individuals with CP and family group only.

Consensus meeting: In Chapter 10, the final stage of COS development methodology was described. The aim of this chapter was to finalise the core outcome (domains) set, which addressed the project objective 1-D. An online consensus meeting was held with key stakeholder groups that contained 21 participants who represented 5 countries. Participants considered the importance of the shortlisted outcomes for COS inclusion, as informed by the results of the Delphi surveys. The importance of outcomes was discussed in small- and large group discussions and voting was conducted in an anonymised manner. The panel reached a consensus that 19 outcomes within eight domains were important and that one outcome was core. These domains were pain, discomfort and fatigue, lower limb structure, motor function, daily life activities, mobility (gait-related outcomes), physical activity, independence, and quality of life. It was recommended that adverse events as a consequence of surgery should always be documented in future studies.

12.2.2 Core Outcome Measurements Set

In order to apply the COS domains effectively, outcomes should be assessed by robust, appropriate and standardised outcome measures. The recommended outcome measurements were selected as the most optimal of the available ones and were defined in a three-stage process, as described in Chapter 11. The three stages are summarised below. This part addressed the second aim of the project.

Definition and evaluation of outcomes measures

Scoping review: An updated review highlighted the significant heterogeneity in outcome measures (n=29) that had been used across 44 studies within three years. Since the completion of the original review (2015), 10 published studies (23%) had sought to use PROMs. The majority of outcome measures addressed impairment of body structure and function (n=20, 69%). Eight outcome measures were used to assess children's activity and participation level. No single outcome measure was used across all studies.

As this review was limited to the most recent outcome measures, the original review findings by Wilson et al. (6) were also included. Duplicate outcome measures were removed and exclusion criteria were applied. Twenty potential outcome measures were shortlisted for inclusion in the core outcome measures set. This finding addressed the project objective 2-A.

Quality assessment: The nine psychometric properties of outcome measures under consideration were assessed using defined criteria: (1) that they were compiled with the COSMIN four-points checklist, which was drawn up to evaluate the methodological quality of measurement properties; and (2) that the studies met quality guidelines in that the researchers had evaluated the reported findings of the outcome measures psychometric properties. The findings from the assessment of points 1 and 2 contributed to best evidence synthesis. The feasibility aspects of the outcome measures were also identified. This stage addressed the project objective 2-B.

Achievement of consensus on core outcome measurement set

Consensus meeting: This meeting formed the final stage of development of the core outcome measurement set. An online consensus meeting was held with seven expert healthcare professionals and researchers who represented two countries. Participants discussed the feasibility of using the outcome measures and highlighted concerns regarding some aspects of the psychometric properties of the identified outcome measures.

After consideration of both the quality and the feasibility of the measurements, eight outcome measures were identified as the most appropriate among the available ones for the assessment of the COS domain. The outcome measures that were recommended by the consensus group were a combination of clinician-driven measures (objective); 3-DGA, EVGS, 10-MWT and GMFM, as well as patient-reported outcome measures (PROMs); GOAL, Gillette FAQ, PROMIS and CP-QoL. These outcome measures were

considered to be of robust quality, and practical and feasible to use. This stage addressed the final objective of the project (objective 2-C).

12.3 Implications and contributions

The work presented in this thesis has made valuable contributions to the knowledge in this field. It has significant implications in research and clinical practice that involves the measurement of outcomes in lower limb orthopaedic surgery for children with ambulant CP.

First, the scoping review identified the outcomes of lower limb orthopaedic surgery that were most reported in the 2016-2019 period. As discussed, the review highlighted important heterogeneity in the reported outcomes and outcome measures. This heterogeneity raised concerns regarding research waste. Whilst wide variation in findings was expected, it was surprising that the most frequently reported outcome domains in previous literature overlapped very little with those that reached consensus among health professionals, children with CP and their families, as the final COS. For example, quality of life outcomes were rarely assessed and independence measures were not collected during previous research in this field, yet they were identified through the interviews and consensus process of this study as important for the stakeholders. This highlights that the outcomes most frequently measured by researchers are not necessarily relevant to or valued by clinicians or by children and young adults with CP or their parents. Future researchers would be advised to involve consultation with key stakeholders in the selection of the primary research outcomes and should consider the proposed COS to minimise research waste.

Second, the identified outcome domains that reached consensus as important demonstrated that individuals with CP and their parents valued lower limb orthopaedic surgery for its benefits on body structure by reducing pain and fatigue, increasing muscle power and joint mobility and improving lower limb symmetry and alignment. However, they also valued the surgery for its potential functional benefits related to decreased occurrence of falls, increased mobility, greater ability to take part in physical activity and increased levels of independence. This indicates the importance of assessing the result of the surgical intervention beyond its potential benefits to the body. Participants agreed that the benefits from the surgery should not be focused on the structural outcomes alone. Instead, its functional benefits in terms of maintaining physical activity levels, enhancing gait-related outcomes, and ultimately improving quality of life should be emphasised.

There is a need for a shift from clinician-reported retrospective outcomes to the use of validated assessment tools that measure patient-reported outcomes, especially related to quality of life and independence. Additional outcomes beyond those that make up the proposed COS may be evaluated, but the proposed COS is recommended as a minimum dataset. Exceptions can be made, for instance, if researchers propose to study a wide range of individuals with CP such as ambulant and non-ambulant patients, or those who are classified within GMFCS I-V.

Third, this work contributed new knowledge through exploring patient and healthcare factors that may impact on the success of lower limb surgical interventions. It is the first study to provide insight into these factors in the context of these surgical interventions. The findings may assist researchers and clinicians to identify and address healthcare system and personal factors, thus promoting surgical 'good outcomes' for patients. These factors also play a key role in the process of obtaining informed consent for surgery.

Finally, although the main aim is to recommend COS to be used in future research studies, the outcomes of importance could also inform clinical practice, particularly when seeking informed consent for a surgical procedure. However, it is not expected that all core outcomes will be routinely collected in clinical practice.

12.4 Recommendations

Several recommendations arose from the findings of this DPhil project and are discussed below.

Recommendation 1: Future researchers assessing the results of lower limb orthopaedic surgery and post-surgical rehabilitation in children with CP should consider evaluating all of the outcome domains agreed in the COS presented in this thesis, using the most optimal measurement tools, as described above. Assessments should be undertaken both before and after surgical interventions in order to evaluate the outcome.

Recommendation 2: Healthcare professionals, children and parents would be advised to consider the differences between key goals and outcomes that can be measured. The terms 'goal' and 'outcome' are often used interchangeably, but they are not the same thing. *Goals* are individualised and specific; they

are statements that reflect the desired outcomes or aim of care for that individual. They should be realistic and written with the patient. Goals could be described in terms of steps to be taken to reach health outcomes. *Outcomes* are expected changes in a patient's health status as a results of an intervention, and they depend on the clinician's knowledge of clinical information. Outcomes could be described as the end results of an intervention to be achieved (569).

Recommendation 3: Those outcome measures that fail to meet threshold scientific quality are not recommended for use in clinical practice and research until high-quality studies for their psychometric properties provide more robust evidence to determine whether they are fit for purpose or not.

Recommendation 4: Issues such as the cost of licenses to use an outcome measures and the additional training needs for staff require to be addressed. This could be achieved through education on the application and interpretation of outcome measures and, most importantly, through offering realistic solutions related to availability of resources and time.

Recommendation 5: Development of a national and international implementation plan is vital and should be carried out with the involvement of key stakeholder groups to ensure that the outcomes and COSs (domain and measures) that are collected and used are relevant and useful.

12.5 Strengths and Limitations

The strengths and limitations of each part of the study were discussed within the relevant chapters. Some broader strengths and limitations of the COS study are discussed below.

Strengths

Patient and Public Involvement: PPI was a key methodological component of the study and was considered a strength of the COS project. Patients and the broader public (parents) were involved as a research partner throughout the study development, including consideration of how the interviews would be structured, topic guide development, the Delphi survey piloting and the consensus and steering groups.

International representation: The project was based on international representation in the consensus stage. The project sought to establish data collection sites in each one of the world regions as defined

by the WHO (570). Sites were established in seven regions: the UK, Europe, the US, Canada, Australia, New Zealand, and South Africa; these areas represented four of the six world regions as defined by the WHO. The incorporation of views from an international perspective is crucial since it enhances international acceptability of the COS. Although efforts were made to capture international perspectives, it is unclear if the identified outcomes would be applicable equivalent across cultures. Delphi survey participants were encouraged to contribute any missing outcome domains.

Identification of 'long list' of outcomes: The choice of potential outcomes for COS inclusion was based on multiple sources. Several outcome domains that were identified as important at the consensus stage would not have been identified from a review of outcomes reporting or interviews with key stakeholders alone. For example, independence, which was identified from the qualitative study, reached a high level of consensus in both the Delphi survey and the consensus meeting with key stakeholder groups.

Size of the Core Outcome Set: The number of outcomes contained in the proposed COS is manageable and of acceptable size for use in the field of lower limb orthopaedic surgery. Although there is no clear guidance regarding the ideal number of core outcomes or domains within a COS, it has been suggested that large COS face challenges with implementation. In a study to develop a COS for patients receiving anti-tumour necrosis factor-alpha therapy for ankylosing spondylitis, Zochling et al. described seven outcome domains as "small enough to be practical, but inclusive enough to manage" (571). The OMERACT initiative suggested that COS developers should aim for COSs that comprise no more than nine outcome domains (299).

Core Outcome Set characteristics: Viehweger et al. (142) emphasised the importance of investigating the outcomes over diverse areas of the ICF and quality of life in paediatric orthopaedic studies. This is in line with the proposed COS, which includes outcome domains that cover every area of the ICF-CY framework and beyond. The use of the ICF-CY framework to classify outcomes was a strength of this study as it provided a common language in the field of CP. The COMET initiative recommends that researchers should not choose to classify outcome domains that are limited to one core area (10).

Adherence to reporting guidelines: The study adhered to internationally recognised methodological and reporting guidelines at all stages. These guidelines included the COMET (Chapter 5), ENTREQ (Chapter 6), PRISMA-ScR (Chapter 7), COREQ (Chapter 8), COS-STAR (Chapter 9 and Chapter 10) and COSMIN

guideline (Chapter 11). Adherence to these standards was important in order to enhance the quality and transparency of reporting.

Limitations

Language restriction: This project was conducted in English only. Due to logistical implications, the qualitative study was conducted with participants from one tertiary Hospital only (UK). It is important to acknowledge that patient and family experiences of care and outcomes of importance are likely to be different regionally and internationally, reflecting cultural and healthcare system differences. Although the Delphi study broadened the collection of stakeholders' views internationally, it was dominated by English-speaking representatives, as it was a requirement that participants had sufficient understanding of English to complete the survey. However, the international language of science is English and thus, the language barrier may not have affected the health professionals' group. There was involvement from healthcare professionals from countries where English is not the primary spoken language: these included France, the Netherlands, South Africa, Saudi Arabia, India and Brazil. Recent guidance was published on the optimisation of international representation in Delphi studies after the inception of the present COS (483).

Stakeholder representation: The development of this COS captured the views of multi-stakeholder groups, including healthcare professionals with diverse experiences, researchers with a variety of expertise, individuals with CP and parents of children with CP. However, other potential stakeholders did not participate in the development of this COS, such as funders, government regulatory authorities, healthcare policy groups, trial managers and journal editors and reviewers. However, some of the healthcare professionals and researchers who took part in the consensus development had multiple roles within the field of CP research. For example, one clinical researcher had a role as a journal editor and played a role in the development of clinical guidelines.

12.6 Dissemination

Publication in relevant journals: A protocol paper with details of the methodologies that were considered during the development of the COSs (Chapter 5) has been published in *BMJ Open* (295). The Qualitative Evidence Synthesis (Chapter 6) has been published in the *Journal of Children's Orthopaedics* (307). The Scoping Review (Chapter 7) has been published in *Developmental Medicine and Child Neurology* (227). Parts of the Qualitative study (Chapter 8) have been published in *Health Expectations*

(516), and *Disability and Rehabilitation* (425). The consensus procedure including the Delphi process (Chapter 9) and Consensus meeting (Chapter 10) have been published in *Developmental Medicine and Child Neurology* (465). Plans are in place to publish the core outcome measurements set (Chapter 11).

Presentation at relevant conferences: Dissemination of the specific studies on the qualitative evidence synthesis, the scoping review, the interviews with stakeholder groups and the achievement of the consensus methods have been presented in both posters (Appendix 16) and oral presentations at national and international conferences of the British Paediatric Neurology Association (2021), World Physiotherapy Congress (2021), Council for Allied Health Professions Research (2021), the British Society for Children's Orthopaedic Surgery (2022), and the American Academy for Cerebral Palsy and Developmental Medicine (2022).

Editorial or commentary: High-impact journals such as *Gait and Posture* and *Journal of Children's Orthopaedics* will be contacted to submit editorials or commentaries about the COS. A commentary on our proposed core outcome domains set has been published in *Developmental Medicine and Child Neurology* (572).

Contact with relevant organisations/charities: A summary of findings in the form of motion graphics (animation) (https://www.youtube.com/watch?v=ReBuT4OFy8g) that was developed with the PPI group will be circulated and disseminated via relevant organisations and charities through their websites, formal and informal meetings and social media. The aim is to increase patients' and parents' awareness and recognition of the importance of COS development and implementation.

12.7 Future directions

When to measure?

To achieve homogeneity and transparency in outcome reporting, we should consider defining 'when' to assess the core outcomes. This is crucial work required after the development of a COS. The COMET initiative guideline places minimum focus on this area. The guideline clearly states that 'what' to measure should be developed first, followed by 'how' to measure (10), yet there is only a vague suggestion that 'when' to measure the core outcomes should be considered during the development of COS.

The time when surgical outcomes are measured is important, since the interpretation and relevance of outcomes change over time. The scoping review of outcomes that have been reported across studies identified a wide range of time point assessments from short- to long-term following surgery. During the interviews, the stakeholders identified a wide range of outcomes and described how their experiences and expectations had altered over time. In addition to this, in the Delphi survey, healthcare professionals explained that it was difficult to score the importance of outcomes as their importance changed over time and depended on the duration of recovery.

In this project, it was planned to consider the optimal time to measure core outcomes during the final consensus meeting with the experts to selecting the optimal outcome measurements. However, the meeting setting was changed from face-to-face to online due to the pandemic. As a result of the restricted time of online meetings, the meeting agenda was reduced. Therefore, further work is required to consider the optimal time point at which measurements should be made. This will add value to the COS implementation and uptake.

Impact of surgery on identified outcomes

The effect of lower limb orthopaedic surgery on some of the core outcomes, such as quality of life and independence is not clear. These outcomes are probably determined by multiple parameters, many of which are not affected by surgery. Further quantitative research is required to assess the impact of surgery on the child's level of independence and quality of life. In addition, further qualitative research is required to gain deeper understanding of how improved independence is perceived by those who live with CP.

Implementation and uptake

An important part of future research would be to evaluate the uptake of the COS by primary studies that are published after the publication of this core set. This would assess the impact of this work in avoiding research waste (573). There is no guidance on how to ensure successful implementation and uptake of a COS. A literature or systematic review could be used to monitor if the outcomes within the COS are collected in primary studies of lower limb orthopaedic surgery for children with ambulant children with CP.

Validation and refinement

Although the COS was developed with international multi-stakeholder groups, the majority were English language speakers. Therefore, further work would be required to establish the validity and international applicability of the COS. Panel meetings of experts and conferences or workshops could be used to assess the applicability of the COS to different international settings. Further research would also be helpful in replicating our work in other countries to determine whether the identified COS is valid across different cultural and language contexts.

Periodic review of the COS of the domains will be necessary to ensure that the included outcomes remain important to stakeholders, relevant to current clinical practice and reflect advances in healthcare. New outcomes may be required and a process to consider new outcomes will be necessary. Furthermore, a review of the COS of the measurements will also be necessary as a result of the evolving evidence base and the continued validation/update of existing outcome measures, for example, sensitivity to change of an outcome measure. The COS of measurements recommended in this thesis represents a consensus recommendation on the best tools currently available to measure the COS of domains. However, both the COS domains and the COS measurements should be seen as recommendations to researchers on the basis of the best current evidence. Progress and evolution on those recommendation is expected in the future, both for the domains and their measurement tools. Hopefully, the work described in this thesis represents a solid base for future research in this field.

Outcome measure specification

Although the COS for the outcome measures was developed based on the current evidence related to the existing measures and their ability to measure the identified core outcome domains set, further work is required to specify which elements of these measures is of relevance to the stakeholders. For example, 3DGA was one of the recommended measures, and kinematic and kinetic parameters and their derivatives in the form of gait scores, such as GPS and GDI, are the most commonly used gait measurements. Further research is needed to determine the most relevant gait parameters derived from 3DGA and the key findings of clinical examinations as outcome measures. The importance of good quality gait data to ensure valid and reliable output was also discussed above.

Barriers and facilitators

Barriers to implementation of the COS should be identified. An anticipated barrier is that it may prove difficult to administer and measure all of the outcomes. Further qualitative studies with researchers in

the field to identify barriers to using the COS and to explore solutions may be helpful. Developing new, faster and simpler but adequately validated outcome measures for the COS domains may also facilitate uptake.

12.8 Conclusion

Research on lower limb orthopaedic surgery for children with CP has grown in recent years. However, the evidence produced has not been accepted conclusively. Considerable heterogeneity in the reporting of outcomes has been one of the main barriers in producing convincing evidence through clinical research in this field. The research work undertaken for this thesis has addressed this issue through the development of a COS domains. This reflects outcomes from surgery that are meaningful and relevant to children with CP, their parents and health professionals. Researchers and clinicians are encouraged to report on the COS domains that have been identified in this thesis, which comprises the eight domains of:

1. Pain, discomfort and fatigue,
2. Lower limb structure,
3. Motor function,
4. Mobility in terms of daily life activities,
5. Mobility in terms of walking,
6. Ability to take part in physical activity,
7. Independence, and
8. Quality of Life.

Reviews of the literature indicated that a large number of different outcome measures are available for use following surgery and highlighted the need for outcome measurements standardisation and development of a core outcome measurements set. This research also addressed this issue and identified the most optimal tools currently available to measure the eight core outcomes. The eight outcome measures identified were:

1. Gait Outcomes Assessment List (GOAL),
2. Patient-Reported Outcomes Instruments System (PROMIS),
3. Gross Motor Functional Measure (GMFM-66),
4. 10-Meter Walk Test (10-MWT),

5. Three-Dimensional Gait Analysis (3-DGA),
6. Edinburgh Visual Gait Score (EVGS),
7. Gillette Functional Assessment Questionnaire (Gillette FAQ), and
8. Cerebral Palsy- Quality of Life (CP-QoL).

Adoption of the COS domains and the optimal tools to measure them in future research should lead to more robust evidence on the effectiveness of lower limb orthopaedic surgery for ambulant children with CP. This should in turn, increase the relevance, efficiency, consistency and transparency of research outcomes and ultimately inform clinical practice and improvements in patient care.

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Appendices

Appendix 1 Steering committee: terms of reference





Development of a Core Outcomes Set for Lower-Limb Orthopaedic Surgery in Ambulant Children and Young People with Cerebral Palsy

Steering Committee Terms of Reference

16th September 2019

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Dr Francine Toye
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Background

Musculoskeletal deformity and gait abnormality are common feature in ambulatory cerebral palsy. Lower limb orthopaedic surgery is considered standard treatment with aim in reducing and preventing long-term damage. Currently, a diverse range of outcomes is being used with little consistency between studies, which results in difficult, sometimes impossible, to synthesise results of studies in systematic review and meta-analysis. Most current research on cerebral palsy does not take into account children and young people' expectations and needs, result in important outcomes being omitted from clinical trials.

Reviews in the outcome measures within the scope of lower limb surgery for cerebral palsy revealed a large selection of diverse measures, many with questionable validity, reliability, and responsiveness. Reviews found that around 65% of the trials did not report in detail on adverse events following surgery and there is a trend toward reporting favourable outcomes.

Under these circumstances, a core outcome set (COS) will be proposed to enhance the efficiency, relevance, transparency, and overall quality of cerebral palsy lower limb orthopaedic surgery research.

Purpose of the project

The aim of the project is to develop core outcomes set relevant to clinical and research practice in lower-limb orthopaedic surgery for ambulant children and young people with cerebral palsy. This will be achieved by:

1. Reviewing the literature on the stakeholders' experiences and expectations,
2. Reviewing the outcome measures have been used in the previous literature of the lower-limb orthopaedic surgery,
3. Interview with the stakeholders to identify their priorities and key health outcomes,
4. Two Delphi survey to reach consensus on the important outcomes to be included in the core outcome sets
5. Consensus meeting with the members of the steering committee to generate the final set of the COS,
6. Based on the identified COS domains of health outcomes, core outcome measures will be developed.

Purpose of the committee

The purpose of the steering committee is to provide oversight in the development of the core outcome set and advise throughout the development process. The Steering Committee will assist with generating the final output of important outcome measures.

Responsibilities

- To attend committee meetings. These will be held at **Nuffield Orthopaedic Centre** and will last approximately **one** hour. Connecting remotely will be possible.
- Committee members will be able to contact the research team before and after each steering committee and to have any questions answered during the meetings.
- To read material sent prior to each meeting. This may include short research reports and meeting agenda
- Steering Committee members will be expected to bring their diverse skills, knowledge and expertise to the discussions based on their experience and perspective.
- The primary functions of the Steering Committee by project phase are to:
    - Comment on preliminary results from Literature Review and interview phases
    - Review key outcomes definitions for the 1st round Delphi survey
    - Refining uncertainties arising from 1st round of the Delphi survey
    - Contribute to the final core outcome set prioritisation.

Working method

- **Two to three** meetings will be held throughout the course of one year, organised by the Nuffield Department of Orthopaedics, Rheumatology and Musculoskeletal Science, University of Oxford.
- Topics for the agenda and a short report will be generated by a member of the COS research team.
- Documents will be circulated, by email, at least two weeks in advance of meetings.
- Meetings will include discussions to share thoughts.
- Minutes from the meetings will be written and distributed.

Appendix 2 Scoping search

Initially, the Embase and PROSPERO databases were searched for existing QESs (Table A). Embase is a biomedical literature database and PROSPERO is the database of prospective registered systematic reviews in health and social care. CP terms in conjunction with a qualitative synthesis filter, as suggested by Toye et al., (574) were used in the search of Embase. Only studies that had been written in English had employed human participants and were available in full-text were included. The term 'cerebral palsy' was used to guide the search of the PROSPERO database, with filter restrictions imposed on health area and review type.

Table A Scoping searching finding

Database	Terms	Search limits	Date	Retrieved
EMBASE	(exp cerebral palsy/) AND (qualitative systematic review* OR (systematic review AND qualitative) OR evidence synthesis OR realist synthesis OR (qualitative AND synthesis) OR meta-synthesis* OR meta synthesis* OR meta synthesis OR meta-ethnograph* OR metaethnograph* OR meta ethnograph* OR meta-study OR meta study OR meta study)	- English Language - Human only - Full text	Dec 2017	23
PROSPERO	Cerebral Palsy	- Health area review: Child health, Musculoskeletal, Surgery, Physiotherapy, Rehabilitation - Type and method of the review: Systematic Review, Meta-Analysis, Qualitative Synthesis	Dec 2017	31

Of the total of 54 articles (23 from Embase; 31 from PROSPERO) that were identified through the search, eight reviews were identified (362, 363, 575-580). A summary of the characteristics of the identified reviews van be found in Table B. Four of these reviews had not synthesised primary qualitative research (577-580), and two reviews were outsides the scope of this study as they reported severe, non-ambulatory CP (575, 576). Two reviews had some relevance (362, 363).

Kruijsen-Terpstra et al. (363) published a mixed-method review that included eight qualitative studies in order to understand the experiences of parents of young children with CP in relation to the physical and occupational therapy that the children received in rehabilitation settings. The review reflected only the experience of the parents. Several studies have emphasised the importance of focusing on the child's perspective (581). Kruijsen-

Terpstra et al. included a range of condition severities. Nevertheless, it is important to recognise subgroups within the CP population based on the severity of the condition and the functional ability of the members of these groups, as the healthcare delivery system can be different for those with different levels of severity and functional ability, for example, the needs of ambulatory and non-ambulatory patients are likely to be different.

In 2016, Lindsay published a QES that was based on 33 studies, which aimed to synthesise the experiences and perspectives of children and young people (from 2 to 25 years old) who were living with CP in order to inform the development of rehabilitation and social programmes (362). The synthesis found that young people with CP were affected by pain, fatigue, and impairments to body function as well as by social exclusion. This review did not incorporate the view of multiple viewpoints, which is essential to gain an in-depth understanding of the condition.

Table B Excluded reviews

Author (year)	Aim	Study design	Number of included studies	Analytic method	Analytic output
Bania et al (2011)	Determine if habitual physical activity could be increased in people with cerebral palsy	Mixed studies	5		Descriptive
Bouwhuis et al (2015)	Overview on the effectiveness of preventive and corrective surgical interventions to treat hip luxation in patients with severe cerebral palsy	Observational	15	Best evidence synthesis	2 categories- Descriptive
Figueiredo et al (2008)	Evaluating the quality of current research on the influence of ankle-foot orthoses (AFOs) on gait in children with cerebral palsy	Qualitative	20	Narrative summaries	Descriptive
Keeratisiroj et al (2016)	Investigate the prognostic predictors for ambulation in children with cerebral palsy using meta-analysis of observational studies	Mixed studies	20	Multivariable analysis	Descriptive
Kruijsen-Terpstra et al (2014)	Give an overview of the experiences and related factors of parents of young children with CP in relation to the physical and/or occupational therapy	Mixed studies	13	Framework synthesis	3 categories- 14 themes
Lindsay (2016)	Synthesize the experiences and perspectives of youth living with CP to inform the development of rehabilitation and social programmes	Qualitative	33	Thematic	7 themes
Morgan et al (2014)	Identify, appraise, and synthesize the evidence describing gait decline in adults with cerebral palsy (CP)	Qualitative	16		4 categories- Descriptive
O'Brien et al (2016)	Establishing the current evidence base for physical activity and exercise interventions that promote health, fitness and well- being, rather than specific functional improvements, for children who use wheelchairs	Mixed studies	31		Descriptive

Appendix 3 Qualitative synthesis search terms and results

	Key term search	EMBASE	Medline	PubMed	CINAHL	PsycINFO
1	cerebral palsy.mp. or exp *cerebral palsy/	35533	21961	25920	8695	7083
2	("qualitative research*" or "qualitative study*" or "qualitative method*" or qualitative).mp.	247402	158281	191685	39707	139201
3	(interview* or "focus group*").mp.	427713	307272	342619	199320	307278
4	2 OR 3	593200	407764	463377	229211	432654
5	ambulatory.mp.	136893	137355	73726	28668	7074
6	1 AND 5	1095	649	685	282	123
7	(parent* or caregiver* or "care giver*" or family* or carer*).mp.	1499123	1173563	1420574	254310	517093
8	1 and 7	5143	2541	3220	1472	1624
9	(exp *transition to adult care/ or transition.mp./or transition to adulthood.mp.) AND "cerebral palsy".mp.	295	123	176	97*	77*
10	4 AND 9**	68	30	37	30	17
11	(orthop?edic surgery*/ or p?ediatric surgery*/ or exp *surgery/) and "cerebral palsy".mp.	5948*	1643*	4287*	628*	209*
12	(physiotherapy or "physical therapy" or rehab*) and "cerebral palsy").mp.	7170	2800	7746	2256	962
13	4 AND 8**	792	381	460	359	344
14	1 AND 4**	1503	740	851	683	624
15	6 AND 4**	54	19	22	22	13
16	11 AND 4**	108	39	65	27	10
17	12 AND 4**	486	158	475	240	87
18	(exp *"quality of life"/ or quality of life.mp.) and "cerebral palsy".mp.	1867	828	1045	516	355
19	18 AND 4**	194	97	119	77	50
20	(evaluat* or needs or concer* or satisfaction* or dissatisfaction* or perception* or experience* or expectation* or perceive* or perspective* or understanding or acceptab*).mp.	7169679	4973268	5528360	1101029	2052671
21	1 OR 8 OR 11 OR 12	35552	21961	25923	8698	7042
22	21 AND 20 AND 4**	1503	528	605	559	624

* Search run for each keyword, then combined them
** used for further screening

Appendix 4 Distribution of the studies supporting each ICF-CY concept and each of the two additional themes

Table A Concepts identified from a priori framework (ICF-CY)

ICF-CY code	*Definition*	*Studies*
Body Functions		
B2 Sensory Functions and Pain		
b280	Sensation of pain	(209-211, 361, 581, 582)
B4 Functions of the cardiovascular and respiratory systems		
B455	Exercise tolerance functions (Fatiguability)	(209, 211)
B7 Neuromusculoskeletal and Movement-related functions		
b710	Mobility of joint functions	(209, 210, 581)
b730	Muscle power functions	(211)
b760	Control of voluntary movement functions	(209, 210)
b770	Gait pattern functions	(210, 581)
b780	Sensations related to muscles and movement functions	(210)
Body Structures		
S7 Structures related to movement		
s750	Structure of lower extremities	(210, 581, 582)
Activity and Participation		
D4 Mobility		
d410	Changing basic body position	(211)
d415	Maintaining a body position	(211, 581)
d450	Walking	(209-211, 581, 582)
d455	Moving around	(211, 581)
d460	Moving around in different locations	(210, 211, 581)
D9 Community, social and Civil Life		
d910	Community life	(209, 211)
d920	Recreation and leisure	(210, 211)
Environmental Factors		
E3 Support and Relationship		
e310	Immediate family	(210)
e320	Friends	(210)
e325	Acquaintances, peers, colleagues, neighbors and community members	(210)
e355	Health professionals	(209-211, 361)
E4 Attitude		

e425	Individual attitudes of acquaintances, peers, colleagues, neighbors and community members	(209, 211)
E5 Services, Systems and Policies		
e580	Health services, systems and policies	(211)
e585	Education and training services, systems and policies	(209, 211)
Personal Factors		
Motivation		
Past experience	Intrinsic or self-motivation can be defined as motivation arising from an individual's internal desires for the satisfaction and fulfilment of specific needs	(209, 361)
Complications		(209, 211, 582)
Future outcome		(210)
Unpredict result/poor outcomes		(211, 581)

Table B Concepts identified from thematic synthesis (not covered by ICF-CY)

Code	***Category title***	***Studies***
Emotions and well-being		
Emotions and well-being		
Stress, tiredness, frustration	How individuals feel about their life	(361)
Overwhelming		(361)
Vulnerable		(361)
Goal setting		
Goal-setting roles	The decisions individuals make about setting goals during their treatment sessions	(209-211)

Appendix 5 Scoping review search terms

Database	Search terms
Medline	1. cerebral palsy.mp. or exp*cerebral palsy/ 2. surgical procedure*.mp. 3. surgery.mp. 4. operative.mp. or exp*surgical procedures, operative/ 5. 1 AND (2 OR 3 OR 4)
Embase	1. cerebral palsy.mp. or exp*cerebral palsy/ 2. surgical procedure*.mp. 3. surgery.mp. or exp*surgery/ 4. operative.mp 5. 1 AND (2 OR 3 OR 4)
CINAHL	1. (MM ”cerebral palsy”) or “cerebral palsy” 2. “surgical procedure*” 3. surgery 4. (MM”surgery, operative”) or “operative” 5. 1 AND (2 OR 3 OR 4)
PubMed	1. “cerebral palsy” 2. “surgical procedure*” 3. surgery 4. operative 5. 1 AND (2 OR 3 OR 4)
Cochrane	1. “cerebral palsy” 2. “surgical procedure*” 3. surgery 4. “operative” or “operation surgery” 5. 1 AND (2 OR 3 OR 4)

Appendix 6 Included studies in the scoping review

Author	Study Objectives	Country of Origin	Number of patients	GMFCS	Age	Gender M:F	Method of recruitment	Type of OR	Follow-up period
2016									
de Morais Filho et al	Evaluate whether distal rectus femoris transfer (DRFT) is related to postoperative increase of knee flexion during the stance phase in cerebral palsy (CP)	Brazil	308	I, II, III	Mean A) 10.3 B) 12.6	193:115	Retrospective	SEMLS (RFT)	G1:3.4 y G2: 3.3 y
Saglam et al	Analyse the mid-term results of femoral derotational osteotomy (FDO) on the clinical findings, temporospatial and kinematic parameters of gait in children with CP	Turkey	93		6.2 ± 3.1	198:116	Retrospective	SEMLS (FDO)	1 year
Mallet et al	Assess the impact of IMPL as a component of single-event multi-level surgery (SEMLS) on spatialand temporal gait parameters, clinical hip flexion deformity, and hip flexion kinematics	France	34	II, III	13±2.8	26;8	Retrospective	SEMLS (Intramuscular Psoas lengthening)	2.4±2 years
Švehlík et al	Long-term out- comes of SEMLS in children with bilateral spastic CP	Austria	39	II, III	mean 10 yr 4 months	22;17	Retrospective	SEMLS	10 years
McMulkin et al	Evaluate the gait and functional outcomes of a group of subjects with CP who underwent surgical intervention that included an FDO compared with a match group with indications of internal hip rotation that did not receive an FDO.	USA	133	I, II, III	4.2-20.8	65;68	Retrospective	SEMLS (± FDO)	11.9-13.8 months
2017									
Boyer et al	Examine the effect of femoral derotation osteotomy (FDO) on dimensionless hip abductor moment during gait in children with cerebral palsy.	USA	140	I, II, III	4.5 y - 17.5 y	77: 63	Retrospective	FDO	1 years 3 years
Klotz et al	Analyze the influence of DFESO on knee and pelvis in children with CP.	Germany	22	I, II, III	12.1±3.1 Y	14:08	Retrospective	SEMLS (FDO)	Mean 15.6 months
Schranz et al	Examin the long-term outcome of unilateral SEMLS in children with unilateral spastic cerebral palsy	Austria	14	I, II	6 - 17 YEARS		Retrospective	SEMLS	1 year 3-5 years 10 years

Bohm et al	Which CP patients are at risk for excessive anterior pelvic tilt following correction of flexed knee gait including patellar tendon shortening	Germany	32	II	8-18 Y		Retrospective	SEMLS (PTS, FDO)	24.1 months
Ancillao et al	(i) study gait changes, by means of synthetic descriptors, in children with CP that underwent Single Event Multilevel Surgery; (ii) compare the MAP and the LFM on these patients; (iii) design a new index that may overcome the limitations of the previous methods	Amsterdam, NL	9	II,III	11.1 ± 2.4 years	7;2	Retrospective	SEMLS (HAM)	14.4±4.5 months
Lung et al	Analyse the efficacy of the double transfer as part of multilevel surgery for individuals with combined flexed knee and stiff knee problem during walking.	Hong Kong	6	II, III	11 - 19 YEARS	2;4	Retrospective	SEMLS (RFT, HAM)	2 years
Desailly et al	Assessed the effects of patella lowering as a component of multilevel surgery.	France	24	I, II, III	10.8±1.9 years		Retrospective	SEMLS (PTS)	2.25±0.8 years
Sung et al	Evaluate the outcomes after DHL and analyse the factors that influence the improvement and serial change in knee motion after surgery in patients with cerebral palsy (CP), using a linear mixed model (LMM)	Korea	314	I, II, III	3.4 -20 YEARS	198;116	Retrospective	HAM	2.7±2.9 years
Mansour et al	Determine the efficacy and safety of percutaneous medial hamstring myofascial lengthening	Lebanon	18		7 -12 YEARS	8:10	Prospective (RCT)	SEMLS (HAM)	-
Kläusler et al	Analyse the results of the long-term follow-up and to determine the relapse rate of tibialis anterior tendon shortening (TATS) and Achilles tendon lengthening (ATL)	Switzerland	23	I, II	14.9 ± 4.0 years	13;7	Retrospective	Tibialis Anterior Shortening (TATS) + Achilles Tendon Lengthening (TAL)	1-2 years 3 years
Sousa et al	Compare knee kinematic outcomes in patients with CP and stiff knee gait who underwent single-event multilevel surgery with and without DRFT.	USA	74	I, II, III, IV	8.4 (2.3)	42; 32	Retrospective	SEMLS (RFS)	20-37 months
Bayhan et al	Evaluate the outcomes of non-arthrodesis surgical treatment of hallux valgus (HV) deformity in children with cerebral palsy	USA	25	I, II, III	15±2.8	22;17	Retrospective	HV correction	14.6 months

Aiona et al	Compare the differences between doing DRFT concomitantly with HSL and doing delayed DRFT after HSL.	Australia	111	I, II, III	mean 11.6	74;37	Retrospective	RFL, RFT, HAM)	1 year
2018									
Klotz et al	Investigate if patellar tendon shortening (PTS) as a part of SEMLS (single event multilevel surgery) is effective for reduction of flexed knee gait in children with cerebral palsy (CP) and, if PTS leads to stiff knee gait.	Germany	20	II, III	5- 18 YEAR		RCT	SEMLS (PTS)	12.7 ± 1.6 m onth
de Morais Filho et al	Compare three different approaches used for the treatment of crouch gait in CP [distal femur extension osteotomy (DFEO), patellar tendon shortening (PTS) and the combination of DFEO +PTS] regarding the increase of APT after the interventions.	Brazil	95	I, II, III	A) 12.8 B)15.7		Retrospective	SEMLS (PTS, FDO)	2.6-2.8 years
Dreher et al	Report the long- term outcomes of MLS on gait function at skeletal maturity, investigated the long-term efficacy and safety of multilevel surgery (MLS) in ambulatory children with bilateral spastic cerebral palsy (CP).	Europe/ Australia	231	I, II, III	7:8 - 14.2 YEARS		Retrospective	SEMLS	1.1 ± 0.4 year 9.1 ± 3.0 year
Sung et al	Evaluate the long-term outcomes at more than 10 years after FDO in children with CP	Korea	34	I, II	4.9 - 17.1 YEAR	19:15	Retrospective	SEMLS (RFT, FDO, ADD, HAM)	10.0–21.0 years
El-Hilaly et al	Assessing short term results Calcaneo-cuboid-cuneiform (triple C) osteotomies	Egypt	12	I, II, III, IV	5.1–15.3 years	7;5	Prospective (case series)		10 - 26 weeks
Kim et al	Quantify changes in gait parameters after FDO	Korea	19	I, II	4 - 20 YEARS	10;9	Retrospective	SEMLS (FDO)	1 year
Corradin et al	Investigate the modification of gait kinematic after epiphysiodesis for limb equalization and secondary to verify the prediction of correction.	Italy	10		10-16 years	7; 3	Retrospective		Mean 6.7, range 3-10 years
Lee et al	Evaluate the influence of surgery involving tendons around the knee on ankle motion during gait in the sagittal plane in cerebral palsy patients.	Korea	35	I, II, III	mean 11.2 ± 4.7 years	18;16	Retrospective	RFT, HAM	Mean 2.2 (SD 1.5) range 0.9-6 years
Niklasch et al	Evaluation of the long-term development of limbs with initial overcorrection after FDO.	Germany	64	I, II, III	10.2 (+/2 2.9 years)		Retrospective	SEMLS (HAM)	At least 5 years
Salami et al	Assesses changes in hamstring muscle tendon length (MTL) and velocity after DFEO (+PTA).	Germany	19		Mean 13 YEARs		Retrospective	SEMLS	1 year 2-5 years

Braatz et al	Does FDO lead to a change of internal transversal hip moments? Are preoperative internal transversal hip moments a predictor for recurrence of IRG?	Germany	41	I, II	10.4 ± 2.7		Retrospective	SEMLS (FDO)	At least 5 years
Niklasch et al	Verify, with a larger number of patients, that there is neither a kinematic nor structural difference after a proximal or distal FDO in children with bilateral CP.	Germany, USA	119	I, II, III	9.4 prox FDO, 11.10 distal FDO		Retrospective	SEMLS (FDO)	9-24 months
Steppacher et al	Investigate changes in kinematic and kinetic patterns from preoperative to postoperative assessments in single-event multilevel surgery and between patients undergoing single-event multilevel surgery and healthy controls.	Swizerland	24	I, II, III	CP 14.9 (3.2)		Retrospective	SEMLS	Mean 1.67(SD 20) year
Church et al	Evaluate the long-term outcome of adolescents with cerebral palsy who have undergone SEMLS for a flexed-knee gait,	USA	59	I, II, III, IV	Mean 14 years (SD 2.5)		Retrospective	SEMLS	Mean 12ys (SD 2)
Ellington et al	Compare short-term outcomes of 2 surgical procedures (RFT) and Rectus Intramuscular Lengthening.	USA	46	I, II, III	lengthening 10.8 (6.5-18.2) transfer 10.7 (6.5 - 18.8)	38;8	Retrospective		1.2 year
Nicholson et al	Examined the relationship between the Gait Deviation Index (GDI) and walking activity preoperatively and postoperatively.	USA	74	I, II, III, IV	11 (SD 4)	31;43	Retrospective		10-26 months
Bittmann et al	Investigated relation- ships between patella position, knee flexion, and the patellar tendon moment arm in children treated with the DFEO and PTA procedures.	USA	40		13.7 ± 3 years	29;11	Retrospective	SEMLS (DFEO, PTA)	6 - 36 months
Aboelenein et al	Evaluate the operative management of pes planovalgus deformity in ambulatory cerebral palsy (CP) children by calcaneal lengthening osteotomy described by Evans.	Kuwait	15	I,II	8 .4 -14.6 y	5;10	Prospective	Calcaneal lengthening	31 mon
De Morais Filho	Compare semitendinosus transfer to distal femur (STTX) to semitendinosus surgical lengthening (STL) regarding the increase of anterior pelvic tilt after flexed knee gait treatment	Brazil	39	I, II, III	G1: 10 years + 7 months/ G2: 11 years + 4 months	29;10	Retrospective	semitendinosus transfer to distal femur (STTX) to semitendi	Mean 5 years + 11 months Mean 7 years + 2 months

								nosus surgical lengthening (STL)	
De Morais Filho	Evaluate the effect of the Majestro-Frost soft tissue procedure (MFP), which potentially addresses muscle imbalance, on IHR in CP patients during walking.	Brazil	78	I, II, III	G1: 9y+8m/ G2: 9y+7m	49;29	Retrospective	MFP	21.6 - 34.7 m
Pilloni et al	Investigate the spatiotemporal, kinematic and kinetic parameters of gait, objectively assessed, in a cohort of children who underwent atl to verify the existence of possible strategies to minimize the negative consequences of the crouch gait pattern.	Italy	18	I,II	Mean 9.6±4.7 years	13;5	Retrospective	ATL	12.1±1.5 months
Salami et al	To evaluate short-term (1y postoperatively; E1) and long-term (at least 4y postoperatively; E2) changes in hamstring muscle-tendon length (MTL) and lengthening velocity after hamstring lengthening in children with bilateral cerebral palsy (CP).	Germany	19		6-10 Y	16;3	Retrospective	SEMLS	Mean 13 (SD 3) months Mean 6y (SD 1y)
Saleh et al	Evaluated the outcomes of tendon Achilles lengthening in 12 children (mean age: 11.2 years) with spastic hemiplegia	Canada	12	I,II	11.2 (4.1)	6;6	Prospective	ATL	6 months 12 months 24 months
Dequeker et al	Investigate the evolution in self-care and functional mobility over the first 2 years after SEMLS and to assess whether determinants that have an influence on these time trends could be identified.	Belgium	34	I, II, III	MEAN 12 years 4 months	22;12	Prospective	SEMLS	2 years
2019									
Osborne et al	Determine whether the hamstrings are short and/or slow preoperatively only in patients with primary, and not repeat, HSL.	USA	33	I, II, III	Control: 10.6 (2.1); HSL: 8.6 (2.4); Repeated HSL: 12.6 (1.2)	20;13	Retrospective	HSL	Mean 1.4 ±1.1
Lashkouski et al	Assess rotational reinsertion of lateral layers of the Achilles tendon, benefits by comparing plantar pressure distribution patterns in	Europe	22		9.1 to 14.5	15;7	Retrospective	Rotational Reinsertion of the	6 -12 mon

	children preoperatively and at 6- and 12-month intervals postoperatively							Lateral Layers of the Achilles Tendons	

NDORMS
NUFFIELD DEPARTMENT OF ORTHOPAEDICS, RHEUMATOLOGY AND MUSCULOSKELETAL SCIENCES

UNIVERSITY OF OXFORD

Development of a core outcomes set for surgical orthopaedic interventions in ambulant children and Young People (CYP) with Cerebral Palsy



VOLUNTEERS NEEDED FOR INTERVIEW

To help patients, doctors and other health professionals make decisions about treatments, we need evidence about what works best. At the moment, different studies looking at treatments for the same condition often measure different outcomes. Hence, we cannot compare or combine their results because they have used different outcomes. **The aim of the study is to develop an agreed set of main outcomes taking into account the views of patients, their families and the health care professionals.**

We invite Parents/carers of (8-18 years old) with cerebral palsy (who can walk independently or with support)

You will take part in a single **60-minute interview** session.

You would be asked to share your perspective on the key health issues that should be measured after orthopaedic treatment in cerebral palsy

If you are interested and would like more information, please contact the researcher **Hajar Almoajil** at the Botnar Research Centre, Headington, Oxford on [07474515165] or **hajar.almoajil@ndorms.ox.ac.uk**.
We will offer gift vouchers to participants and will cover travel expenses.

Version/Date: 1.1 (29/07/2019)
Chief Investigator: Mr. Tim Theologis

IRAS Project number: 260093
REC Reference number: 19//SC/03

NDORMS
NUFFIELD DEPARTMENT OF ORTHOPAEDICS, RHEUMATOLOGY AND MUSCULOSKELETAL SCIENCES

UNIVERSITY OF OXFORD

We can see you at **HOME** or **HOSPITAL**

60 minutes Chat and Play

Are you diagnosed with **CEREBRAL PALSY**?

Are you able to **walk with or without assistance**?

Are you **8 - 18 years old**?

Please we need your help

hajar.almoajil@ndorms.ox.ac.uk

[07474515165]

There is **£20 voucher** for participants

Talk to your **family**, and contact me

Version/Date: 1.1 (29/07/2019)
IRAS Project number: 260093
REC Reference number: 19/SC/03
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Appendix 8 Interview topic guides

Core Outcome Set (Interview Topic Guide)
Healthcare Professionals

Introduction

- Go over the purpose of the study with participant.
- Check they are still willing to take part.
- Check they are happy for interview to be audio recorded.
- Answer any other queries.
- Ask them to sign the consent form.

Interview Themes/Questions

General Question

1. What are some of your experiences with children and young people (CYP) with cerebral palsy throughout the time you have cared for them? These can apply to any age group for any musculoskeletal treatment for their lower limb/to improve their gait.

Probe
- Based on experience, what are some considerations you typically have with these patients? And or their carers

Assume participants will provide some experiences they have with this population

General experiences on outcomes

2. Overall, what lower-limb orthopaedic aspects of CYP's health matter most to you when working with this population, specifically for their lower-limb deformity?

Assume participants will provide some unique outcomes or considerations for this group

3. What are concerns or experiences that CYP/carers typically share with you post-surgery?

Probe
- This may include topics that are related to the CYP and/or family/carer, on physical health, daily routine or quality of life, etc

Assume participants will share some concerns that raised by patients and carers

Specific Questions on outcomes

4. Are there any differences between what you and the CYP/carers deem important?

Follow up questions:
- What might be the causes of any of these differences?

Expect discussions on outcomes that they considered important, where not relevant or important to the patients/carer

5. Which considerations do you have that you think other professionals/carer/patients over- emphasize, or under-emphasize?

Assume a discussion of general outcomes, in terms of what should be appreciated more or less

6. What are the five outcomes you value most when it comes to lower-limb surgery of CYP with cerebral palsy, and should definitely be measured in the research area? and why?

Expect valued outcomes and discussion

7. Moving to the systematic review results, outcomes that are reported in the literature include: [example of outcomes]. Do you have any opinions on any of these lists in general?

Assume discussion on some outcomes from the systematic review results

Closing questions

8. To sum up, after discussing and listing various perspectives and outcomes, could you provide me with two or three outcomes that you believe essential to consider?

Expect most valued outcomes list

9. Is there anything else you would like to add?

Closing up interview

- Thank you for participating.

Core Outcome Set (Interview Topic Guide)
Parent/carer

Introduction

- Go over the purpose of the study with participant.
- Check they are still willing to take part.
- Check they are happy for interview to be audio recorded.
- Answer any other queries.
- Ask them to sign the consent form.

Interview Themes/Questions

General Question

1. I understand you have a child with cerebral palsy, please could you describe some experiences about how your life has been affected? Are there any experiences, which have significantly changed the way you live as an individual, or in terms of family, friends?

Follow up question
- What has the treatment of your child's leg problems been like?

Assume participants will provide some experiences they have as parent/carer of a child with cerebral palsy with this condition and treatment, to stimulate conversation on the topic in general.

General experiences on outcomes

2. Overall in terms of your child's health, what matters most to you when considering orthopaedic surgery for his/her leg problem(s)?

Assume participants will provide some of main priorities

3. Is there anything that you have been thinking about related to your child's leg surgery that you feel hasn't been addressed yet in your discussions with the treating team?

Probe
- Are there any outcomes/issues/concerns you expected treatment to address but have not been considered?

Assume either no input, or some examples of outcomes/concerns not been addressed.

Specific Questions on outcomes

4. When you discussed the results of your treatment with the team that treated your child, were there any issues where you didn't agree with them? Were there aspects of the treatment that you considered important and they did not, or the other way round?

Probe:
- Which aspects do you think have been over-emphasised?
- Was there anything that needs to be more emphasised?

Follow up questions:
- What do you think might be the cause of any of these differences?

Expect outcomes that they deem important, that healthcare professionals may not consider.

5. When you think of the treatment for your child's leg problems through surgery, what are the most important results you would wish them to achieve?

Follow up question
- Why do you think these results are important?
- Has what you consider important changed over time?

Expect list of valued outcomes and subsequent discussion

6. Outcomes that are reported from other young people with your child's condition include: [example of outcomes], what are your thoughts on those?

Follow up question
- Have you discussed any of these with your doctor?

Assume discussion on some outcomes from the systematic review results

Closing questions

7. To sum up, after discussing and listing various aspects of results and outcomes, could you provide me with two or three main issues that you think essential to consider?

Expect most valued outcomes list

8. Is there anything else you would like to add?

Assume either no input for this question, or further discussion

Closing up interview

- Thank you for participating.
- Providing participant £20 voucher and sign the voucher receipt.

Core Outcome Set (Interview Topic Guide)
Children and Young People (16 -1 8 years)

Introduction

- Make sure the accessibility for the child mobility is available [e.g., accessible entrance and adjusted seat].
- Go over the purpose of the study with participant.
- Check they are still willing to take part.
- Check they are happy for interview to be audio recorded.
- Prompt for and answer any other queries.
- Ask them to fill in the consent form.

Interview Themes/Questions

General Question

1. I understand you have cerebral palsy; please could you describe some experiences about how your life has been affected? Are there any experiences, which have significantly changed the way you live as an individual, or in terms of family, friends?

Follow up question
- What has the treatment of your leg problems been like?

Assume participants will provide some experiences they have with this condition and treatment, to stimulate conversation on the topic in general.

General experiences on outcomes

2. As you progress towards your adulthood, are there any more considerations, concerns or different experiences that arise for you?

Follow up question
For outcomes that may emerge:
- Which of these have been the most important to you?

Assume participants will provide some highlighted experiences and share some outcomes that may have emerged.

3. What are the main pieces of advice you would share with a friend, who has a similar condition, particularly about going through leg surgery?

Potential Probe
- We're interested in hearing about your expectations from the treatment, the care you need, and any issues related to your health or well-being?

Assume participants will share major experiences and considerations.

Specific Questions on outcomes

4. When you discussed the results of your treatment with the team that treated you, were there any issues where you didn't agree with them? Were there aspects of your treatment that you considered important and they did not, or the other way round?

Probe:
- Which aspects do you think have been over-emphasised?
- Was there anything that needs to be more emphasised?

Follow up questions:
- What do you think might be the cause of any of these differences?

Expect outcomes that they deem important, that healthcare professionals may not consider.

5. When you think of the treatment for your leg problems through surgery, what are the most important results you would wish to achieve?

Follow up question
- Why do you think these results are important?
- Has what you consider important changed over time?

Expect list of valued outcomes and subsequent discussion

6. Outcomes that are reported from other young people with your condition include: [example of outcomes], what are your thoughts on those?

Follow up question
- Have you discussed any of these with your doctor?

Assume discussion on some outcomes from the systematic review results

Closing questions

7. To sum up, after discussing and listing various aspects of results and outcomes, could you provide me with two or three main issues that you think essential to consider?

Expect most valued outcomes list

8. Is there anything else you would like to add?

Assume either no input for this question, or further discussion

Closing up interview

- Thank you for participating.
- Providing participant £20 voucher and sign the voucher receipt.

Core Outcome Set (Interview Topic Guide)
Children and Young People (8 -15 years)

Introduction

- Make sure the accessibility for the child mobility is available [e.g., accessible entrance and adjusted seat].
- Go over the purpose of the study with participant.
- Check they are still willing to take part.
- Check they are happy for the interview to be audio recorded.
- Prompt for and answer any other queries.
- Ask the parent to fill in the consent form and the child complete the age appropriate assent form.

The questions asked will be based on the talking mat tools **(talking mat example, version 01, date 25 /07/2019)**. As such these questions are possible examples of the type of questions that will be asked. Therefore, these questions will not be used precisely as they are and may be modified depending on the outcome of the activity, patient age and understanding.

Interview Themes/Questions

General Question

1. Introduce to talking mat

2. How do you feel about [different cards]? Within a scale of "important, unsure, not important" pictures, table A and B.

Follow up question
- is there anything else that is not here, but is important to you?

Assume participants will share their experiences and discuss outcomes that may have emerged.

Different question about it:
How do you think about your mobility around home, school... etc? How do you feel about ...?

Table A: Questions with different activities

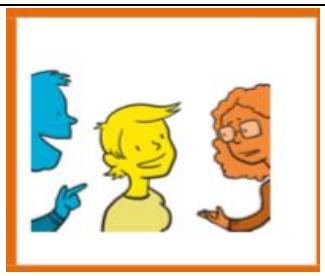
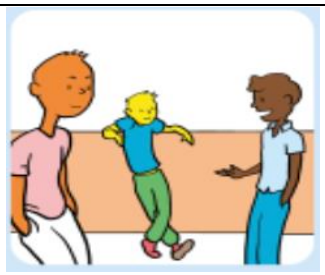
Using technologies	
Socialising with friend/family	
Participation with peers	
Bicycle (sport)	

Table B: Scale

		
Assume different activities will be considered as important.	Assume different activities will be considered as unsure.	Assume different activities will be considered as not important to participants.



An example of a mat for the expected results will be on the same pattern

General experiences on outcomes

3. Choose which symbols you like the best [from important group]? From the completed activity above (Q2).

Follow up question
- Why do you like these activities?

Assume participants will provide some activities that are important to them.

4. How do you think about [different activities shown in cards]? These items were identified from someone else in your age or a family member, and/or doctors.

Potential Probe
- Of all cards you find important, are there any that your family/ doctors also find important?
- Can you tell me why?

Expect outcomes that they deem not important to them, that healthcare professional, family members may consider as important.

Specific Questions on outcomes

5. Are there any activities that you used to do but no longer doing because of your leg problem?

6. Did the surgery fix these problems and you able to do it now? Or Do you expect your future leg surgery will fix these problems and you will become able to do these activities?

Assume discussion over their experience with the condition and related surgery

7. What are the most important results [from cards] you would wish to achieve?

Follow up question
- Why do you think these results are important?

Expect list of valued outcomes and subsequent discussion

8. How do you feel about how long things took to be achieved after your leg surgery?

Assume participants will be share some experience about goals achievement.

Closing questions

9. To sum up, could you pick up two or three main things that you feel it is important to achieve after you had leg surgery?

Expect most valued outcomes list

10. Is there anything else you would like to talk about? Closing up interview

Closing up interview

- Thank you for participating.
- Providing participant £20 voucher and sign the voucher receipt.

Appendix 9 Short-listed outcomes

To decrease the child's pain and muscle soreness
To feel less pain
To feel less muscle soreness or tiredness after activities
To improve the way the child looks
To be able to stand taller or more upright
To have straighter and better shaped legs
To have legs that look the same as each other
To increase how much the child can do
To reduce or prevent falls
To be able to climb stairs
To be able to do more activities at home
To improve participation in sports (e.g., football, swimming)
To improve participation in hobbies (e.g., dancing, horse-riding)
To increase the child's independence
To be able to go out of the house independently (without help from an adult)
To be able to do things independently at home
To be able to do activities of daily living independently (e.g., bathing, dressing and self-care)
To be able to walk without assistance when going out
To be able to walk without assistance at home
To improve the way the child walks after surgery
To be able to walk with straight legs
To be able to walk without getting tired
To be able to walk faster
To be able to walk further
To be able to walk without using a crutch or walker
To be able to walk better even if continue using a crutch or walker
To stop wearing a brace /splint/ orthosis
To improve how the child feel when interacts with other people
The child improves friendships and relationships with other people
To feel more confident
To feel happier
To feel better about how other people treat the child
Concerns about having surgery
Feeling pain after the surgery
Developing wound infection
Losing independence
Needing to have another surgery
Getting worse after surgery
Taking a long time to walk again
How important are the following factors in your decision to recommend the surgery?
Good access to physiotherapy services locally
Good communication with other healthcare professionals
Being well informed about the operation and potential risks
Getting help from the right health professionals when needed
Getting support from family/friends

Appendix 10 National and international organisations and charities

A) Healthcare professionals

Name	Country
The British Society for Children's Orthopaedic Surgery (BSCOS)	UK
Action Cerebral Palsy	UK
Cerebral Palsy Society	New Zealand
The Pediatric Orthopaedic Society of North America (POSNA)	USA
The European Paediatric Orthopaedic Society (EPOS)	
The Paediatric Orthopaedic Society of India	India
The South African Paediatric Orthopaedic Society (SAPOS)	South Africa
The Australian Paediatric Orthopaedic Society (APOS)	Australia
The Association of Paediatric Charted Physiotherapists	UK
The Middle East Paediatric Orthopaedic Society (MEPOS)	Middle East
International Cerebral Palsy Society	
Kids Brian Health Network	Canada
The American Academy for Cerebral Palsy and Developmental Medicine (AACPDM)	worldwide

B) Children and Young People with CP, Families

Name	Country
Action Cerebral Palsy	UK
Cerebral Palsy	UK
SCOPE	UK
International Cerebral Palsy Society	
CP Potential	UK
Cerebral Palsy Society	New Zealand
Cerebral Palsy Foundation	USA
Cerebral Palsy Scotland	UK
STEP	UK
United Cerebral Palsy	USA and Canada
Childhood Disability Link	Canada
The South African Paediatric Orthopaedic Society (SAPOS)	South Africa

Appendix 11 Consensus meeting discussion summary

PAIN

- To feel less pain

Although the group believed that pain was under-reported in clinical trials for ambulatory children with CP, they agreed that it was important and should be included in the COS. The discussion also revealed that the term was clearly understood, and several pain-related questions should be asked to acquire a complete picture of pain related to CP. One surgeon descried this as *"the first question is do you have pain anywhere and the second question is where is it and the third is offer them a Likert scale of 1 to 9 to know the severity of pain and the frequency of pain"*.

FATIGUE

- To feel less muscle soreness or tiredness after activities

Like pain, the group recognised that outcomes related to fatigue that have significant impact on patients and their family are poorly recorded and reported. However, they felt that it's a huge a problem to the children and their family life and that it was important to measure and should be included in the COS. During the discussion, the group felt there was ambiguity around fatigue because they imply this includes **(I) muscle fatigue** and **(II) energy level after activities or tiredness.** They believed that the concept of fatigue was and underrated by clinician. For example, using three-Dimensional Gait Analyse (3-DGA), which is a standard tool to assess the surgical interventions outcome, is a source of fatigue as it exceeds the child's capacity. Thus, they recommended that clinician may look at this issue and ask the question *"how far the kids really can go to complement our gait laboratory measurement?"*.

LOWER LIMB STRUCTURES

- To have straighter and better shaped legs

- To have legs that look the same as each other

The research team agreed that outcomes were belong to two inter-related goals, namely (I) lower limb alignment, and (II) lower limb symmetry. Participants agreed with these inter-related goals, however, they agreed there is lack of clarity of definition for many of the terms used to describe these outcomes, particularly between clinicians and non-clinicians. This justified the difficulties to reach consensus on the Delphi survey results. The group has taken the decision to alter the purpose of these surgical goals to (I) aesthetic purpose (i.e., mechanical axes and alignment in different planes) and (II) functional purpose (e.g., impact on gait). The group agreed that the lower limb alignment during activity that may impacted the child's life, rather than the aesthetic should be used when measuring lower limb structure as the latter could mirror the functional purpose of improving better lower limb alignment. Though, measuring lower limb symmetry for both aesthetic purpose (make them less different from other kids) and functional purpose are considered important.

STANDING POSTURE

- To be able to stand taller or more upright

The terms upright and taller were discussed. The group felt there was ambiguity around these terms and were felt to be reported separately. The term 'standing taller' was not felt to be accurate or meaningful clinically or biologically, and they agreed that it was not impotent and should be drop out from the COS. On the other hand, the group felt 'standing upright' was agreed was important when capture and facilitate functional aspect of health (i.e., during walking) as one parent stated

"I think about it I don't only think of standing but also probably walking when you're able to stand more upright while walking. You don't have to spend as much time looking down to make sure that you clear from any potential obstacles". Therefore, standing position for functionals purpose should be included in the COS. The group felt that it's impossible to separate standing up right from achieving better lower limb alignments.

STANDING BALANCE AND FALL

- To reduce or prevent falls

- To be able to stand with a better balance

Participants agreed that standing balance is a huge contributor to falling prevention, so grouping them together was acceptable and they are very important aspect of surgical goals. They also agreed that lower limb alignment and symmetry play an important role to prevention of fall incident.

MOBILITY: INDOOR MOBILITY

- To be able to climb stairs

- To be able to do more activities at home

Mobility indoor include the ability to climb stair and doing activity indoor such as self-care. The group agreed that overall outcomes specific indoor mobility was clearly understood terms and they important as captured real-world environment outcomes. Although the group were emphasised on the importance of the ability of climb a stair as source of safety, they felt it is how accessible is the environment to the children for disability rather than a surgical impact. Participants also agreed that climbing a stair require other element such alignment and flexibility.

MOBILITY: WALKING

- To be able to walk with straight legs

- To be able to walk with the foot flat on the ground

- To be able to walk without getting tired

- To be able to walk faster

- To be able to walk further

- To be able to walk without using a crutch or walker

- To be able to walk better even if continue using a crutch or walker

- To stop wearing a brace /splint/ orthosis

The walking related outcomes include: (I) walking pattern (lower limb straight and foot flat on the ground), (II) walking endurance, (III) walking distance, and (IV) walking speed. The four outcomes listed within this category were thought to be important and representative of how these outcomes have been described in available outcome measures and to include all the key areas where patients' lives may be impacted. However, there was one of the more involved discussions of the day as there is lack of clarity of the term used to describe 'walking speed', which reflecting the lack of consensus in the Delphi survey. The group agreed to rephrase this outcome toward **(I) Maintain walking speed compared to presurgical status** and **(II) Walking speed to keep up with others**.

Other construct related to walking is using assistive device, comprise: (I) orthosis, splint or brace, and (II) crutch or walker. The group agreed that the decision to use or not to use of orthosis, splint or brace were clearly controversial. The participants

also agreed that there are several reasons behind not use them such as aesthetic issue. On the other hand, they noted that there are some benefits of using the orthosis, splint or brace as they contributes to the child's stability, walking speed and keep up with others, as a parent of a child stated, *"the decision was made based on the speed of her walk so she's so much faster with them she actually now wears them three times a week so that she can keep up with friends at school so I think that just goes to show how important"*. Overall, agreement was reached that the concept of 'to stop using orthosis, splint or brace' should be included as an outcome in the COS despite the fact that for some children, it helps them walking more efficiently.

The group raised two issues related to using crutch or walker, which may justify the Delphi findings around using assistive devices. First, the group has taken the decision to distinguish between the term 'crutch' from 'walker' on the statement since not relying on crutch or walker depends on the child's level of function pre-surgery. A surgeon explained this and supported by a study findings (124) as *"that if you haven't graduated from a Walker to crunches by the time you're leaving the Children's Hospital you're not going to be a community ambulator in in later life you're going to go back to a wheelchair whereas if you've been if you've been able to reach independence with crunches rather than a Walker you have a pretty good chance of maintaining independent mobility"*.

Second, the group felt there was ambiguity around 'walking better' on the statement because they imply this includes walk with a better kinematic pattern or efficiently (i.e., distance and speed). the group suggested to rephrase the outcome to **'To rely less on assistive aid'**. Because health professionals cannot promise or predict changes of the surgery on the use of the assistive aid, the group suggested this outcome should not be part of the COS despite its importance. This could be discussed as part of decision making, a parent of a child reported this as, *"I think it is important as a measure of expectation and how expectations get set but not necessarily a measure of success of surgery"*.

PARTICIPATION

- To improve participation in sports (e.g., football, swimming)

- To improve participation in hobbies (e.g., dancing, horse-riding)

- To be able to join more activities at school (e.g., PE)

This was one of the more involved discussions of the day as there is lack of clarity of definition for many of the terms used to describe these outcomes, for example, participation in sport, hobbies or school activities. It was agreed that listing participation outcomes simply in general terms would be unsatisfactory as it is recognised that particular outcomes that have significant impact on patients were not defined (e.g., physical activity). The group also felt that these outcomes were complex terms as they are subjective to the individual child interest. Therefore, it poses a challenge on how best to assess these outcomes.

The group suggested rephrasing these terms to reflect surgical outcomes in relation to participation domain. **(I) Participation on social activities of interests:** this includes outcomes related to individual or group sport activities (e.g., swimming, football), hobbies (e.g., dancing, horse-riding), school or community activities. Due to subjectivity of this outcomes and the fact the its impacted hugely by postoperative rehabilitation, it was agreed to not included as primary outcomes within the COS, (**II) Maintain physical activity:** the group consensus on the importance of pre and postoperative physical activity and that should be included in the COS. During the discussion, a physiotherapy emphasised on this *"if you're going to provide*

surgery you definitely don't want physical activity to be lease a year afterwards or two years afterwards so I think it is when we already know children with CP are mainly sedentarily so it could be an outcome that is really important".

INDEPENDENCE

- To be able to go out of the house independently (without help from an adult)

- To be able to do things independently at home

- To be able to do activities of daily living independently (e.g., bathing, dressing and self-care)

- To be able to walk without assistance when going out

- To be able to walk without assistance at home

The independence referred to a person's level of functional independence without a support from an adult. The domain includes outcomes related to indoor and outdoor activities, walking independently indoor and outdoor. The outcomes listed within this domain were thought to be representative of how these outcomes meaningful and important to the children and families. Therefore, the group accepted and agreed the independence related outcomes should be included in the COS. Further specification was deemed necessary as it will fall under the remit of rehabilitation postoperative. The group agreed that dialogue around expectation goals play a vital part in surgical decision making. One surgeon explained this as the follow: *"I had seen a girl who had SEMLS at growth spurt secondly deformities who referred back with a full plan to put everything right and when she filled in the GOAL questionnaire nothing was to do with gait everything was to do with independence activities of daily living, and I said OK she's voted no surgery that's got physio and strengthening going and surgeries off the question. so, this is absolutely according with what we get in our lab from the GOAL questionnaire and it's a real insight and a real wake up alert for surgeons"*.

QUALITY OF LIFE

- The child improves friendships and relationships with other people

- To feel more confident

- To feel happier

- To feel better about how other people treat the child

Although the group accepted the term quality of life impact as important outcome, they believed that the surgery cannot impacted the child's quality of life directly and is not a primary goal of surgery. For example, the child's quality of life may improve as consequences of not wearing splint and the walking ability improved after surgery. The outcomes listed on this category were altered as they were felt by the group not to be clinically meaningful and that adapted environment (accessibility) and social education may hugely impacted the child quality of life.

The group also felt that the outcomes of Quality of Life mirrors other functional outcomes considered such as lower limb symmetry, mobility, and participation outcomes and they agreed that the terms **(I) self-esteem**, and **(II) body image** should be used when describing quality of life related to the surgery. This is in keeping with the QOAL questionnaire, which is patient reporting outcome measure for lower lib surgical interventions.

ADVERSE EVENTS

- Feeling pain after the surgery

- Developing wound infection

- Losing independence

- Needing to have another surgery

- Getting worse after surgery

- Taking a long time to walk again

The adverse events include the following items: getting worse than presurgical status, surgical recurrence, losing independence, time of recovery, postsurgical pain syndrome, and infection. The group agreed that all the six outcomes within Adverse Event (AE) are not surgical outcomes, in addition to **skin pressure damage**. The consensus was reached on this and the feeling from the clinicians and patients and the parents in the group was that it was information they would want to see reported in trials, but not required to be included in the COS. The group discussed the importance of reporting the adverse events, this include: (I) evaluation of new treatment, (II) informed decision making, and (III) inform patients and families about associated risk.

Although they felt reporting the adverse events is important, some adverse events do not reflect the success of surgery long term rather considered to be the presence at a specified point in time after surgery such infection. The group agreed that there is lack of clarity of definition for some of the term used to describe some adverse events. For example, (I) surgical recurrence, the group felt it was not clear enough if repeating surgery is due to growth and development factors or relapse of the first operation, and (II) time of recovery, it was not clear if this was impacted by the long-term rehabilitation perception and not surgery alone.

FACTORS IMPACT SURGICAL DECISION MAKING

- Good access to physiotherapy services locally

- Good communication with other healthcare professionals

- Getting help from the right health professionals when needed

- Being well informed about the operation and potential risks

- Getting support from family/friends

- Ability to adhere to home exercises

The group agreed that these factors are important on the surgical success and confirmed that these not surgical outcomes. No firm conclusion was reached on this however the feeling from the clinicians, patients and the parents in the group was that it was information on the factors that may impact surgical outcomes and satisfaction level postoperatively. Physiotherapy availability and information resource of benefit and risk of the surgery were felt to be needed during decision making process. Overall treatment time was felt to be the preferred term to describe radiotherapy compliance.

Appendix 12 Consensus meeting summary table

Domains and Outcomes	Discussion	Final decision
To decrease the child's pain and muscle soreness	Change the domain name to fit with existing outcome measures	Pain, discomfort, and fatigue
To feel less pain	Definitely in	Retain item
To feel less muscle soreness or tiredness after activities	Muscle fatigue Definitely in	Retain items
	Fatigue (tiredness) after activity Definitely in	Retain items
To improve the way the child looks		
To be able to stand taller or more upright	Exclude taller Exclude better shaped leg (better is vague term)	Refine item
To have straighter and better shaped legs	Definitely in	
To have legs that look the same as each other	Change to symmetry Definitely in	Retain item
To increase how much the child can do	Separate motor function, mobility, and participation domain	- Motor function - Mobility: DLA - Participation
To reduce or prevent falls	Definitely in (motor function)	Retain item
To be able to stand with a better balance	Balance impacts the fall incident, and could be together Definitely in (motor function)	
To be able to climb stairs	Unsure, depended on the context, and could be with DLA In (Mobility: DLA)	Refine item (Mobility: DLA)
To be able to do more activities at home	Definitely in (mobility: DLA)	
To improve participation in sports (e.g., football, swimming)	Replaced with social activity or physical activity (participation), depended on the context	Retain item (Physical activity)
To improve participation in hobbies (e.g., dancing, horse-riding)	Definitely in (participation: physical activity)	Exclude (Social activity) depend on individual child own interest
To be able to join more activities at school (e.g., PE)		
To increase the child's independence		
To be able to go out of the house independently (without help from an adult)	Definitely in	Retain item
To be able to do things independently at home To be able to do activities of daily living independently (e.g., bathing, dressing and self-care)	Join these outcomes together Definitely in	Retain item
To be able to walk without assistance when going out	Definitely in	Retain item
To be able to walk without assistance at home	Definitely in	Retain item
To improve the way the child walks after surgery		

To be able to walk with straight legs	Walking pattern Definitely in	Retain item
To be able to walk with the foot flat on the ground	Walking pattern Definitely in	
To be able to walk without getting tired	Definitely in	Retain item
To be able to walk faster	Relaced to maintain walking speed Definitely in	Retain item
To be able to walk further	Definitely in	Retain item
To be able to walk without using a crutch or walker	For patient education and decision making	Refined: to rely less on assistive device Exclude
To be able to walk better even if continue using a crutch or walker	Definitely out	Exclude
To stop wearing a brace /splint/ orthosis	Definitely in	Retain item
To improve how the child feel when interacts with other people	Change domain to Quality of Life	Quality of Life
The child improves friendships and relationships with other people	Definitely out	Exclude
To feel more confident	Related to (self-esteem) Definitely in	Self-esteem Retain item
To feel happier	Definitely out	Exclude
To feel better about how other people treat the child	Related to (body image) Definitely in	Body image Retain item
Concerns about having surgery	Change domain to adverse event	Adverse Event
Feeling pain after the surgery	Definitely in For documentation only	Retain item
Developing wound infection		Retain item
Losing independence		Retain item
Needing to have another surgery		Retain item
Getting worse after surgery		Retain item
Taking a long time to walk again		Retain item
How important are the following factors in your decision to recommend the surgery?		
Good access to physiotherapy services locally	Important factors, but not outcome Definitely out	Exclude
Good communication with other healthcare professionals	Replaced to: Surgeon and Hospital factors, availability of good services	Exclude
Getting help from the right health professionals when needed		
Being well informed about the operation and potential risks	Important factors, but not outcome Definitely out	Exclude
Getting support from family/friends		Exclude
Ability to adhere to home exercises		Exclude

Appendix 13 COSMIN Checklist

Box A. Internal consistency				
	excellent	**good**	**fair**	**poor**
1 Does the scale consist of effect indicators, i.e. is it based on a reflective model?				
Design requirements				
2 Was the percentage of missing items given?	Percentage of missing items described	Percentage of missing items NOT described		
3 Was there a description of how missing items were handled?	Described how missing items were handled	Not described but it can be deduced how missing items were handled	Not clear how missing items were handled	
4 Was the sample size included in the internal consistency analysis adequate?	Adequate sample size (≥100)	Good sample size (50-99)	Moderate sample size (30-49)	Small sample size (<30)
5 Was the unidimensionality of the scale checked? i.e. was factor analysis or IRT model applied?	Factor analysis performed in the study population	Authors refer to another study in which factor analysis was performed in a similar study population	Authors refer to another study in which factor analysis was performed, but not in a similar study population	Factor analysis NOT performed and no reference to another study
6 Was the sample size included in the unidimensionality analysis adequate?	7* #items and ≥100	5* #items and ≥100 OR 6-7* #items but <100	5* #items but <100	<5* #items

7	Was an internal consistency statistic calculated for each (unidimensional) (sub)scale separately?	Internal consistency statistic calculated for each subscale separately			Internal consistency statistic NOT calculated for each subscale separately
8	Were there any important flaws in the design or methods of the study?	No other important methodological flaws in the design or execution of the study		Other minor methodological flaws in the design or execution of the study	Other important methodological flaws in the design or execution of the study
Statistical methods					
9	for Classical Test Theory (CTT), continuous scores: Was Cronbach's alpha calculated?	Cronbach's alpha calculated		Only item-total correlations calculated	No Cronbach's alpha and no item-total correlations calculated
10	for CTT, dichotomous scores: Was Cronbach's alpha or KR-20 calculated?	Cronbach's alpha or KR-20 calculated		Only item-total correlations calculated	No Cronbach's alpha or KR-20 and no item-total correlations calculated
11	for IRT: Was a goodness of fit statistic at a global level calculated? E.g. χ^2, reliability coefficient of estimated latent trait value (index of (subject or item) separation)	Goodness of fit statistic at a global level calculated			Goodness of fit statistic at a global level NOT calculated

Box B. Reliability: relative measures (including test-retest reliability, inter-rater reliability and intra-rater reliability)					
		excellent	**good**	**fair**	**poor**
Design requirements					
1	Was the percentage of missing items given?	Percentage of missing items described	Percentage of missing items NOT described		
2	Was there a description of how missing items were handled?	Described how missing items were handled	Not described but it can be deduced how missing items were handled	Not clear how missing items were handled	
3	Was the sample size included in the analysis adequate?	Adequate sample size (≥100)	Good sample size (50-99)	Moderate sample size (30-49)	Small sample size (<30)
4	Were at least two measurements available?	At least two measurements			Only one measurement
5	Were the administrations independent?	Independent measurements	Assumable that the measurements were independent	Doubtful whether the measurements were independent	measurements NOT independent
6	Was the time interval stated?	Time interval stated		Time interval NOT stated	
7	Were patients stable in the interim period on the construct to be measured?	Patients were stable (evidence provided)	Assumable that patients were stable	Unclear if patients were stable	Patients were NOT stable
8	Was the time interval appropriate?	Time interval appropriate		Doubtful whether time interval was appropriate	Time interval NOT appropriate

9	Were the test conditions similar for both measurements? e.g. type of administration, environment, instructions	Test conditions were similar (evidence provided)	Assumable that test conditions were similar	Unclear if test conditions were similar	Test conditions were NOT similar
10	Were there any important flaws in the design or methods of the study?	No other important methodological flaws in the design or execution of the study		Other minor methodological flaws in the design or execution of the study	Other important methodological flaws in the design or execution of the study
Statistical methods					
11	for continuous scores: Was an intraclass correlation coefficient (ICC) calculated?	ICC calculated and model or formula of the ICC is described	ICC calculated but model or formula of the ICC not described or not optimal. Pearson or Spearman correlation coefficient calculated with evidence provided that no systematic change has occurred	Pearson or Spearman correlation coefficient calculated WITHOUT evidence provided that no systematic change has occurred or WITH evidence that systematic change has occurred	No ICC or Pearson or Spearman correlations calculated
12	for dichotomous/nominal/ordinal scores: Was kappa calculated?	Kappa calculated			Only percentage agreement calculated
13	for ordinal scores: Was a weighted kappa calculated?	Weighted Kappa calculated		Unweighted Kappa calculated	Only percentage agreement calculated
14	for ordinal scores: Was the weighting scheme described? e.g. linear, quadratic	Weighting scheme described	Weighting scheme NOT described		

Box C. Measurement error: absolute measures				
	excellent	**good**	**fair**	**poor**
Design requirements				
1 Was the percentage of missing items given?	Percentage of missing items described	Percentage of missing items NOT described		
2 Was there a description of how missing items were handled?	Described how missing items were handled	Not described but it can be deduced how missing items were handled	Not clear how missing items were handled	
3 Was the sample size included in the analysis adequate?	Adequate sample size (≥100)	Good sample size (50-99)	Moderate sample size (30-49)	Small sample size (<30)
4 Were at least two measurements available?	At least two measurements			Only one measurement
5 Were the administrations independent?	Independent measurements	Assumable that the measurements were independent	Doubtful whether the measurements were independent	measurements NOT independent
6 Was the time interval stated?	Time interval stated		Time interval NOT stated	
7 Were patients stable in the interim period on the construct to be measured?	Patients were stable (evidence provided)	Assumable that patients were stable	Unclear if patients were stable	Patients were NOT stable
8 Was the time interval appropriate?	Time interval appropriate		Doubtful whether time interval was appropriate	Time interval NOT appropriate

9	Were the test conditions similar for both measurements? e.g. type of administration, environment, instructions	Test conditions were similar (evidence provided)	Assumable that test conditions were similar	Unclear if test conditions were similar	Test conditions were NOT similar
10	Were there any important flaws in the design or methods of the study?	No other important methodological flaws in the design or execution of the study		Other minor methodological flaws in the design or execution of the study	Other important methodological flaws in the design or execution of the study
Statistical methods					
11	for CTT: Was the Standard Error of Measurement (SEM), Smallest Detectable Change (SDC) or Limits of Agreement (LoA) calculated?	SEM, SDC, or LoA calculated	Possible to calculate LoA from the data presented		SEM calculated based on Cronbach's alpha, or on SD from another population

Box D. Content validity (including face validity)					
		excellent	**good**	**fair**	**poor**
General requirements					
1	Was there an assessment of whether all items refer to relevant aspects of the construct to be measured?	Assessed if all items refer to relevant aspects of the construct to be measured		Aspects of the construct to be measured poorly described AND this was not taken into consideration	NOT assessed if all items refer to relevant aspects of the construct to be measured

2	Was there an assessment of whether all items are relevant for the study population? (e.g. age, gender, disease characteristics, country, setting)	Assessed if all items are relevant for the study population in adequate sample size (≥10)	Assessed if all items are relevant for the study population in moderate sample size (5-9)	Assessed if all items are relevant for the study population in small sample size (<5)	NOT assessed if all items are relevant for the study population OR target population not involved
3	Was there an assessment of whether all items are relevant for the purpose of the measurement instrument? (discriminative, evaluative, and/or predictive)	Assessed if all items are relevant for the purpose of the application	Purpose of the instrument was not described but assumed	NOT assessed if all items are relevant for the purpose of the application	
4	Was there an assessment of whether all items together comprehensively reflect the construct to be measured?	Assessed if all items together comprehensively reflect the construct to be measured		No theoretical foundation of the construct and this was not taken into consideration	NOT assessed if all items together comprehen-sively reflect the construct to be measured
5	Were there any important flaws in the design or methods of the study?	No other important methodological flaws in the design or execution of the study		Other minor methodological flaws in the design or execution of the study	Other important methodological flaws in the design or execution of the study

Box E. Structural validity				
	excellent	**good**	**fair**	**poor**
1 Does the scale consist of effect indicators, i.e. is it based on a reflective model?				
Design requirements				
2 Was the percentage of missing items given?	Percentage of missing items described	Percentage of missing items NOT described		
3 Was there a description of how missing items were handled?	Described how missing items were handled	Not described but it can be deduced how missing items were handled	Not clear how missing items were handled	
4 Was the sample size included in the analysis adequate?	7* #items and ≥100	5* #items and ≥100 OR 5-7* #items but <100	5* #items but <100	<5* #items
5 Were there any important flaws in the design or methods of the study?	No other important methodological flaws in the design or execution of the study		Other minor methodological flaws in the design or execution of the study (e.g. rotation method not described)	Other important methodological flaws in the design or execution of the study (e.g. inappropriate rotation method)

Statistical methods					
6	for CTT: Was exploratory or confirmatory factor analysis performed?	Exploratory or confirmatory factor analysis performed and type of factor analysis appropriate in view of existing information	Exploratory factor analysis performed while confirmatory would have been more appropriate		No exploratory or confirmatory factor analysis performed
7	for IRT: Were IRT tests for determining the (uni-) dimensionality of the items performed?	IRT test for determining (uni)dimension-ality performed			IRT test for determining (uni)dimension-ality NOT performed

Box F. Hypotheses testing					
		excellent	**good**	**fair**	**Poor**
Design requirements					
1	Was the percentage of missing items given?	Percentage of missing items described	Percentage of missing items NOT described		
2	Was there a description of how missing items were handled?	Described how missing items were handled	Not described but it can be deduced how missing items were handled	Not clear how missing items were handled	
3	Was the sample size included in the analysis adequate?	Adequate sample size (≥100 per analysis)	Good sample size (50-99 per analysis)	Moderate sample size (30-49 per analysis)	Small sample size (<30 per analysis)

4	Were hypotheses regarding correlations or mean differences formulated a priori (i.e. before data collection)?	Multiple hypotheses formulated a priori	Minimal number of hypotheses formulate a priori	Hypotheses vague or not formulated but possible to deduce what was expected	Unclear what was expected
5	Was the expected *direction* of correlations or mean differences included in the hypotheses?	Expected direction of the correlations or differences stated	Expected direction of the correlations or differences NOT stated		
6	Was the expected absolute or relative *magnitude* of correlations or mean differences included in the hypotheses?	Expected magnitude of the correlations or differences stated	Expected magnitude of the correlations or differences NOT stated		
7	for convergent validity: Was an adequate description provided of the comparator instrument(s)?	Adequate description of the constructs measured by the comparator instrument(s)	Adequate description of most of the constructs measured by the comparator instrument(s)	Poor description of the constructs measured by the comparator instrument(s)	NO description of the constructs measured by the comparator instrument(s)
8	for convergent validity: Were the measurement properties of the comparator instrument(s) adequately described?	Adequate measurement properties of the comparator instrument(s) in a population similar to the study population	Adequate measurement properties of the comparator instrument(s) but not sure if these apply to the study population	Some information on measurement properties (or a reference to a study on measurement properties) of the comparator instrument(s) in any study population	No information on the measurement properties of the comparator instrument(s)

9 Were there any important flaws in the design or methods of the study?	No other important methodological flaws in the design or execution of the study		Other minor methodological flaws in the design or execution of the study (e.g. only data presented on a comparison with an instrument that measures another construct)	Other important methodological flaws in the design or execution of the study
Statistical methods				
10 Were design and statistical methods adequate for the hypotheses to be tested?	Statistical methods applied appropriate	Assumable that statistical methods were appropriate, e.g. Pearson correlations applied, but distribution of scores or mean (SD) not presented	Statistical methods applied NOT optimal	Statistical methods applied NOT appropriate

Box G. Cross-cultural validity				
	excellent	**good**	**fair**	**poor**
Design requirements				
1 Was the percentage of missing items given?	Percentage of missing items described	Percentage of missing items NOT described		
2 Was there a description of how missing items were handled?	Described how missing items were handled	Not described but it can be deduced how missing items were handled	Not clear how missing items were handled	

3	Was the sample size included in the analysis adequate?	CTT: 7* #items and ≥100 IRT: ≥200 per group	CTT: 5* #items and ≥100 OR 5-7* #items but <100 IRT: ≥200 in 1 group and 100-199 in 1 group	CTT: 5* #items but <100 IRT: 100-199 per group	CTT: <5* #items IRT: (<100 in 1 or both groups
4	Were both the original language in which the HR-PRO instrument was developed, and the language in which the HR-PRO instrument was translated described?	Both source language and target language described			Source language NOT known
5	Was the expertise of the people involved in the translation process adequately described? e.g. expertise in the disease(s) involved, expertise in the construct to be measured, expertise in both languages	Expertise of the translators described with respect to disease, construct, and language	Expertise of the translators with respect to disease or construct poor or not described	Expertise of the translators with respect to language not described	
6	Did the translators work independently from each other?	Translators worked independent	Assumable that the translators worked independent	Unclear whether translators worked independent	Translators worked NOT independent
7	Were items translated forward and backward?	Multiple forward and multiple backward translations	Multiple forward translations but one backward translation	One forward and one backward translation	Only a forward translation
8	Was there an adequate description of how differences between the original and translated versions were resolved?	Adequate description of how differences between translators were resolved	Poorly or NOT described how differences between translators were resolved		

9	Was the translation reviewed by a committee (e.g. original developers)?	Translation reviewed by a committee (involving other people than the translators, e.g. the original developers)	Translation NOT reviewed by (such) a committee		
10	Was the HR-PRO instrument pre-tested (e.g. cognitive interviews) to check interpretation, cultural relevance of the translation, and ease of comprehension?	Translated instrument pre-tested in the target population	Translated instrument pre-tested, but unclear if this was done in the target population	Translated instrument pre-tested, but NOT in the target population	Translated instrument NOT pre-tested
11	Was the sample used in the pre-test adequately described?	Sample used in the pre-test adequately described		Sample used in the pre-test NOT (adequately) described	
12	Were the samples similar for all characteristics except language and/or cultural background?	Shown that samples were similar for all characteristics except language /culture	Stated (but not shown) that samples were similar for all characteristics except language /culture	Unclear whether samples were similar for all characteristics except language /culture	Samples were NOT similar for all characteristics except language /culture
13	Were there any important flaws in the design or methods of the study?	No other important methodological flaws in the design or execution of the study		Other minor methodological flaws in the design or execution of the study	Other important methodological flaws in the design or execution of the study

Statistical methods		
14 for CTT: Was confirmatory factor analysis performed?	Multiple-group confirmatory factor analysis performed	Multiple-group confirmatory factor analysis NOT performed
15 for IRT: Was differential item function (DIF) between language groups assessed?	DIF between language groups assessed	DIF between language groups NOT assessed

Box H. Criterion validity				
	excellent	**good**	**fair**	**poor**
Design requirements				
1 Was the percentage of missing items given?	Percentage of missing items described	Percentage of missing items NOT described		
2 Was there a description of how missing items were handled?	Described how missing items were handled	Not described but it can be deduced how missing items were handled	Not clear how missing items were handled	
3 Was the sample size included in the analysis adequate?	Adequate sample size (≥100)	Good sample size (50-99)	Moderate sample size (30-49)	Small sample size (<30)
4 Can the criterion used or employed be considered as a reasonable 'gold standard'?	Criterion used can be considered an adequate 'gold standard' (evidence provided)	No evidence provided, but assumable that the criterion used can be considered an adequate 'gold standard'	Unclear whether the criterion used can be considered an adequate 'gold standard'	Criterion used can NOT be considered an adequate 'gold standard'

5	Were there any important flaws in the design or methods of the study?	No other important methodological flaws in the design or execution of the study	Other minor methodological flaws in the design or execution of the study	Other important methodological flaws in the design or execution of the study
Statistical methods				
6	for continuous scores: Were correlations, or the area under the receiver operating curve calculated?	Correlations or AUC calculated		Correlations or AUC NOT calculated
7	for dichotomous scores: Were sensitivity and specificity determined?	Sensitivity and specificity calculated		Sensitivity and specificity NOT calculated

Box I. Responsiveness				
	excellent	**good**	**fair**	**poor**
Design requirements				
1 Was the percentage of missing items given?	Percentage of missing items described	Percentage of missing items NOT described		
2 Was there a description of how missing items were handled?	Described how missing items were handled	Not described but it can be deduced how missing items were handled	Not clear how missing items were handled	
3 Was the sample size included in the analysis adequate?	Adequate sample size (≥100)	Good sample size (50-99)	Moderate sample size (30-49)	Small sample size (<30)
4 Was a longitudinal design with at least two measurement used?	Longitudinal design used			No longitudinal design used
5 Was the time interval stated?	Time interval adequately described			Time interval NOT described

6	If anything occurred in the interim period (e.g. intervention, other relevant events), was it adequately described?	Anything that occurred during the interim period (e.g. treatment) adequately described	Assumable what occurred during the interim period	Unclear or NOT described what occurred during the interim period	
7	Was a proportion of the patients changed (i.e. improvement or deterioration)?	Part of the patients were changed (evidence provided)	NO evidence provided, but assumable that part of the patients were changed	Unclear if part of the patients were changed	Patients were NOT changed
Design requirements for hypotheses testing					
	For constructs for which a gold standard was not available:				
8	Were hypotheses about changes in scores formulated a priori (i.e. before data collection)?	Hypotheses formulated a priori		Hypotheses vague or not formulated but possible to deduce what was expected	Unclear what was expected
9	Was the expected *direction* of correlations or mean differences of the change scores of HR-PRO instruments included in these hypotheses?	Expected direction of the correlations or differences stated	Expected direction of the correlations or differences NOT stated		
10	Were the expected absolute or relative *magnitude* of correlations or mean differences of the change scores of HR-PRO instruments included in these hypotheses?	Expected magnitude of the correlations or differences stated	Expected magnitude of the correlations or differences NOT stated		

11	Was an adequate description provided of the comparator instrument(s)?	Adequate description of the constructs measured by the comparator instrument(s)		Poor description of the constructs measured by the comparator instrument(s)	NO description of the constructs measured by the comparator instrument(s)
12	Were the measurement properties of the comparator instrument(s) adequately described?	Adequate measurement properties of the comparator instrument(s) in a population similar to the study population	Adequate measurement properties of the comparator instrument(s) but not sure if these apply to the study population	Some information on measurement properties (or a reference to a study on measurement properties) of the comparator instrument(s) in any study population	NO information on the measurement properties of the comparator instrument(s)
13	Were there any important flaws in the design or methods of the study?	No other important methodological flaws in the design or execution of the study		Other minor methodological flaws in the design or execution of the study (e.g. only data presented on a comparison with an instrument that measures another construct)	Other important methodological flaws in the design or execution of the study
Statistical methods					
14	Were design and statistical methods adequate for the hypotheses to be tested?	Statistical methods applied appropriate		Statistical methods applied NOT optimal	Statistical methods applied NOT appropriate

Design requirement for comparison to a gold standard					
	For constructs for which a gold standard was available:				
15	Can the criterion for change be considered as a reasonable gold standard?	Criterion used can be considered an adequate 'gold standard' (evidence provided)	No evidence provided, but assumable that the criterion used can be considered an adequate 'gold standard'	Unclear whether the criterion used can be considered an adequate 'gold standard'	Criterion used can NOT be considered an adequate 'gold standard'
16	Were there any important flaws in the design or methods of the study?	No other important methodological flaws in the design or execution of the study		Other minor methodological flaws in the design or execution of the study	Other important methodological flaws in the design or execution of the study
Statistical methods					
17	for continuous scores: Were correlations between change scores, or the area under the Receiver Operator Curve (ROC) curve calculated?	Correlations or Area under the ROC Curve (AUC) calculated			Correlations or AUC NOT calculated
18	for dichotomous scales: Were sensitivity and specificity (changed versus not changed) determined?	Sensitivity and specificity calculated			Sensitivity and specificity NOT calculated

Appendix 14 COSMIN Database: Excluded reviews

Review	Ref
Appraised by different tools	
A systematic review of the psychometric properties of Quality-of-Life measures for school aged children with cerebral palsy	(198)
Did not evaluate relevant outcome measure	
Measuring quality of movement in cerebral palsy: a review of instruments	(583)
Primary psychometric properties study	
A systematic review of utility values in children with cerebral palsy	(584)
Out of the review scope	
Tactile assessment in children with cerebral palsy: a clinometric review	(585)
Structure/ scoping review	
Child or family assessed measures of activity performance and participation for children with cerebral palsy: a structured review	(586)
Outcome measures for Adults with CP	
How to measure pain in neurological conditions? A systematic review of psychometric properties and clinical utility of measurement tools	(587)
Did not specify health condition	
Fatigue in child chronic health conditions: a systematic review of assessment instruments	(588)

Pain, discomfort and fatigue

Do we want to be measuring across different constructs can you say it across different constructs do you mean like pain frequency pain intensity pain interference?

Actually, do that they don't measure across aspects of playing you know there's nothing like the McGill pain questionnaire or something like that they just asked fairly simple questions in the **PedsQL** for example he just all said you have 18 pains but all with us.

I work a little bit with the pain researcher and she doesn't even love like the **PROMIS** for instance and the **GOAL questionnaire** is newer and it's okay I like that they have the different areas the locations of the pain but it doesn't isolate right and left sides and I think that information would be important when you're trying to look at outcomes but if you have two sides that are paying full and maybe one is worse than the other is unclear how their answering that questionnaire parents or child if they're responding for the average side average pain score the worst one.

Some of the questionnaires don't ask location of pain at all and so I just think if we don't have a questionnaire that isolates the location of the pain, I mean I know the **PROMIS**. I think it's hard to use that as an orthopaedic outcome tool. I was going to say the location would be important especially given our population cerebral palsy which it's reasonable to think they could have pain in other places you know this isn't like a sports med clinic where it's likely if you're asking a global question about pain, they're talking about this one specific area that's been injured for example.

I think coming from clinical perspective the **PedsQL** is the one that people have reported being the biggest to fill out so if you in a busy clinical environment that's the shorter questionnaire it was said covers full age range as far as I know and that's the next thing go ahead. The only question about pain is asking bout general there's one single question of pain which is the to summary seems to imply that the other questions are about pain that's all the questions are that impact on sleep but the impact on sleep is actually in the section which says about emotions in fact it's not directly referring cause it

I would say I mean I'm not trying to nominate it but the only thing I would say about it is that it reasonably good psychometric properties if we look at the table that Hajar has produced seems to have a reasonable amount to use not massive but a reasonable amount to use across child health in north Germany it is dare I say it is sure isn't there like a CP module. I don't think we should it must have specifically worded about this pain interfere with your sleep I don't think we should consider that the measuring that construct that just doesn't seem appropriate.

Whether people have used the **GOAL**, I think what's clear with the outcome measures is that there's going to be overlap in terms of an outcome measure may record different outcomes, so I was just wondering to say **GOAL** as Hajar said is kind of the new questionnaire. I just wondered if people had an experience of using it and in practice and what you thought of it. Angela has been using it in our gait lab since May 2018, so we have several hundred individuals who we just used the parent version. I think it is nice because it has the frequency like basically none to everyday or multiple times today and then the intensity of it. I think it is maybe a four-point Likert scale and you know with the exception of not identifying right side versus left side for the different enjoying I think it’s pretty satisfactory. It doesn't go really discuss interference right if you think about like modified **PedsQL** where it talks about interference with sleep or activity or this and that it does not distinguish that so I think having that element of interference in a pain questionnaire is important right cause that might be that deciding factor when people seek treatment it's really interfering with my sleep it's you know like I can tolerate and intensity of two during walking but you know something like that so probably the two main limitations.

I would say physical does target do you want to improve upon this so that may be something that conditions can consider your rate in that it's not measuring how much is this interfering. You rate it frequency and intensity so doesn't specifically asked is interfere with your walking but then you can report whether or not it's an area to improve which I think as a clinician but that could be that missing link potentially because I think the question about do you want this improved covers they shouldn't interfere in interference indirectly if you like it doesn't it very likely another thing to consider with all of these is

you know if you're money to measure pain and whether it's clinically or part of a research study I mean almost everything on that on the list except for **PROMIS** which has the pain interference like Kristen said you can mix and match all of these are in like the context of several measures and so some touch it aside you know if you were measuring if you wanted to measure pain only pain you probably wouldn't take out sub set of the **GOAL** right your so that's another thing is it kind of depends on what the research study is or maybe we're just trying to understand a recommendation for clinical practises or both a lot of these things are within much larger sets which fits well with what we want here because here we want to cover the whole width of outlook and domains that we want to evaluate so we don't mind really the pain scoring is not a stand-alone scoring system and this fault about why the questionnaire so I think unless there any other building remarks.

There might just not be one measure but actually depending on your research question and your setting but there might be a selection of tools stop domains over tool but that a researcher might want to use.

I think a possible criticism would come from the ICHOM who promote health outcomes set, their argument is you should always be using the four or five measures they nominate and then adding in the other stuff. From my experience people pick and choose from what who's been nominated but that is one thinking. I think maybe this is more a PhD issue if you did decide to go down that route which I have great sympathy with, but you have to source address that in the discussion of the thesis I mean that's a different issue.

We use **PROMIS** clinically and we're evaluating whether or not it's appropriate for CP but it's this balance of do we ever want to compare across other populations sometimes the answer is yes or are we only looking for change within a population or discrimination within a population and then that also needs to be taken into account so we started with the **PROMIS** to seek as we wanted to be able to compare and **Gillette FAQ** a lot of kids with disabilities and complex conditions so that's why we went with **PROMIS** and now we're evaluating how sensitive is it.

I think that what you're saying is go running the **PROMIS** in parallel with the **GOAL** so you have measurements of both on a number of children so we will have a subset, so the **GOAL** is collected every patient down in our gait motion analysis lab and then clinically in our orthopaedic practise and our spine practise were collecting it before and after surgery

We would recommend **PROMIS** and you can use whatever questions you like but we would always advocate you use these seven questions so might that be so compromised?

I would argue no, because you know CP so get your right there is the short form which is the you ask these 7/12/15 whatever it is no matter what and then the computer adaptive tests which is where it holds depending on your previous response there are cross box between the two but the challenge was if you ask somebody who can't walk and you're using the pain does it hurt to run a mile or whatever I forget the exact wording are they going to say no coz I don't run so it never hurts or yes if I did run would be awful so this is the challenge given just the variability in our seeking population, I think it would be hard to just recommend a short form.

I was just trying to think of a simple solution but is there a simple solution 45 minutes and has only been talking about pain?

Fatigue: I think that's a big complaint of from CP on the research agenda that CPRN came out with an they should address its important patients either this sufficiently covered by the **GOAL** I'll be honest I don't like how the **GOAL** completes it with pain and that pain domain. I wished it was a separate domain on its own but I think there's so much unknown about the experience of fatigue in these individuals that questionnaire and a lot more research currently.

I think there's only two questions on the **GOAL** in that domain see talking about pain discomfort and fatigue it's feeling tired while walking and feeling tired during other physical activities that he or she usually enjoys the problem.

The **GOAL** has been designed specifically for the condition of interest but if you want to compare the level of disability and functionally children with other conditions than having a more generic measure like the **PROMIS** would be with extremely helpful.

Lower limb structure

We'll start with the **GOAL**, we use it and I think it does a pretty good job of addressing the cosmesis concerns of families you don't have it pulled up but it's basically are your feet flat on the ground or you walking upright and not in Crouch, are your feet straight ahead, walking without dragging his feet and dragging the those before or is it looking taller and more operate and then tripping and falling which you have a balance and falls domain. I think that separate would we say it's in the gait pattern and appearance section. I think they don't have a fall on balance domain.

I will say is often a concern like families report that knees knocking and so that's not asked on the **GOAL.** So, I don't know if you create write your own module if you would just end that one item in for the conditions with would you agree that that's a common concern of some of the families that you treat.

I'm align in the general under the body image section. It talks about after all but it's not supported as part of the other domain. So is the gait in appearance and the body image is about it, talks about shape and position of the legs. It doesn't directly ask the questions posed but that's that something that we come up with the gait analysis as well.

Motor function

GMFM is quietly in the paediatric physio world, because this covers only standing and form.

The **GMFM**, it's quite timely the good thing is that it's good into liability so experience I've had it's been really well used across conditions reliability but it's timely it's an hour to perform last your admin afterwards and actually I think for the child is quite a lot even though it's the shorter version it's actually still quite a lot for them to take part and possibly in orthopaedics it covers so it's covering everything and actually we don't need to know quite so much perhaps for this.

What about you as a physio how much training does it is it something that you could pick up off the shelf as a job in physio or would you feel that if you were doing it as part of a trial you would need to be trained to do that?

I think you should have training in it but it's fairly straight forward especially if you're used to measure in movement function is sure it is actually, I think you just need to be able to do it a few times and you soon get the hang and there's a sort of sequence to it so you can quite quickly work through.

It is quite good because it does have lots of items so it's very exist quite accurate sensitive compared with some of the others. I guess it is to say that the time side of things it is the time with that take longer with a small child that's being non-compliant my question everything takes longer with all the chocolates you probably wouldn't do it much before five years age.

Mobility: Daily Life Activities

The **Gillette FAQ**, I mean I don't like one question about climbing stairs so I would never pull that single question out so fear but it certainly asks the thinking 22 asks about whether daily life activities like stepping up on a curb skating walking backwards turning a corner.

The **Gillette FAQ** there is the 10 points where you just are 10 descriptors where you just select one of those then it has the additional 22 skills so we usually distinguish those two I think one of the things about the 10 items the **Gillette FAQ** walking level as we call it does not necessarily distinguish between assistive device use primarily at those lower levels one through 4 and so when we've compared this to the **GOAL** for instance the **GOAL** asks about level of assistance for but different activities and so I think it's advantageous because we have like yes maybe you can walk in therapy but you have to use a Walker or whatever and that's not clearly distinguished on the **Gillette FAQ** walking level.

In research studies, it's hard to distinguish between somebody who's using assistive device with somebody who's not in some of the questions sometimes that's okay sentence problem well and there's just so much overlap if you look at GMFCS level ones and where they fall like they might fall between 6 and 10 and the twos might fall between 4 and 10 and so therefore it doesn't discriminate as well and so I think I don't think I can fully endorse it compared to some of the better measures that

distinguish and capability when it factors in assistance as devices do have any more daily life. There is another of discontinuous with clear life activities all is going to say another **GOAL** asked said yes so these are the rest of promising **GOAL** feature.

The **GOAL** I think it does well it seems to distinguish between groups, but you get ceiling effects for the higher functioning individuals with CP on the ADLs domain.

In terms of the **PROMIS,** we are using it clinically. The data hasn't fully been analysed yet, it doesn't appear to be a sense super sensitive, but there are just a couple of publications that I was just referencing again that I've looked at the **PROMIS mobility domains** and they were less likely to detect change compared to some of the other legacy measures. So, I would not favoured **PROMIS** over the **GOAL**.

Mobility: Walking

I think if we wanted to truly isolate walking speed the **TUG** is obviously confounded by the transition times both standing up and sitting down in and as you mentioned, you can gather walking speed from gait analysis as well **10 metre walk test** I'm using it currently well I just finished that study but I like the 10 minute walk tests at their fastest speed because you actually measuring capability rather than most gait analysis that performed at self-selected speed of course you know they can be correlated but how you're feeling or your age can really dictate how complete you're walking and it doesn't necessarily reflect their capability so if you want objective measures of that.

The **GOAL** obviously reports you know perceived ability to keep up with others so I I'm tended to think that are important perhaps equally important in assessing gair from all aspects adding that taking into account with the objective centre is critical and it takes a lot and a greeting from our discussions with our patient group is important

I suppose that if it is not **three-dimensional gait lab** alternative, will be one of the validated visual gait scores I think **EVGS** is the Edinburgh visual gait score, **PRS** position I think that we're all happy.

I think it is helpful in eliminating things. The **FIM** I think that is a lot of different dimensions doesn't it around self-care independence toilet in absolute thing so looking to narrow down are not necessarily one I would pick out because it used a lot with post stroke so self-care stick to control transfers that sort of thing so I think we may be gaining a lot of information perhaps we won't be needing for this. There is question about motor as well but lots of other areas too that's only my own experience of it.

It's only the **GOAL** which inquiring about the use of orthotics which is interesting.

The **PODCI**, we've used it for many years. I don't know exactly when it started making you have any clue 2010 but no yeah so for walking ability it's mostly that transfers and basic mobility core scale but that also has other non-walking elements in its sub scale such as putting apparently putting on your coat I'm surprised it doesn't fall in the upper extremity ones. It has questions about standing or sitting in a chair without holding on so again it kind of more that basic balance as well not just The Walking ability.

Physical Activity

Physical activity but there was one I think with the just these are probably our three finalists are going to be the **GOAL** that bold he and the **PROMIS** featuring every plus the **gait analysis** as an objective measure

Independence

The independence referred to the activities and movements. I really like the **GOAL** for this honestly because even for the for the activities of daily living it asks about how much assistance you need for those activities total moderate minimal or

supervised or completely independent and then again for more of the mobility based ones that asks about if you want you're using assisted advice and then it has the specific braces and mobility aids domain on top of that too and that's more like how happy are you with them but I do like the **GOAL** in that regard.

Quality of Life

I personally will be indecisive as the weather **GOAL** covers the domain of quality of life or you need a separate measure for quality of life if you're going to include in this population? the question is do you need to measure quality of life after the lower limb surgery? which is so there are two questions really do you need to measure and is the goal enough?

I think in the UK the expectation now is that you always measure quality of life in all trials. I think probably we do need to nominate this I mean if there's a short form quality of life measures that we could use maybe that would mean indeed the **PedsQL**.

I agree that we need to measure quality of life I think that's been a challenge that historically we've only measured alignments or you know gait as measured by three-dimensional gait analysis this is where medicine is going to be more patient centred. So, I think we have to ask quality of life in terms of how we do it and you know stupid hill Q out that you sure that's the same as the kids QL.

I'll just throw in **PROMIS** here is another option because they do have domains on social health as they have all these short our problem is the forms, he started collecting it during a pandemic so I have no good data because you know can I hang out with my friends will probably not coz about pandemic so I just I my only concern is looking at other measures.

The **GOAL** covers so many domains to then say let's do the **CP-QoL** to get at some social health or mental health but then it has all these other domains as well it just feels like a disconnect you know.

I know we're probably going to end up recommending a group of measures so just seems that's **QoL Inventory** hasn't come up in some of the other areas yet unless I missed it must be different in some important record coz the self-report **CP qol** has 53 items and the proxy report has 66 whereas the **PedsQL** only has 23 a somewhat different.

The main limitation of the **GOAL** in regard to the self-esteem category is that it only asks about the shape and position of your legs and feet the symmetry of legs how you get around compared to others the way others feel about how you get around and how you're treated by others so it's I mean half of them are about body image and acceptance of that rather than the **CP-QoL** is more comprehensive and in general it's not just focusing the way your body on the looks.

There are three self-esteem questions basically on the **GOAL** that don't explicitly call out legs otherwise ported right next to them so they could keep tied up with you thinking about it so it's not completely absent but yeah again depending on the point of your study it may or may not be sufficient call.

It’s possible families could call something out but that's obviously not going to get scored or anything like that so we probably need a **CP- QoL** measure in addition to the **GOAL**.

a quality of life considering something like the **EQ5D child version** it is I'm not very familiar with it but that maybe some that's just very health related quality of life specific that you could use in health economics as opposed to the **CP-QoL** measure that's on the slide which also covers a number of other aspects that may be already covered passed by things like the **GOAL**.

It crossed covers and clear very simply questions and it has to be said that the **EQ5D** measures don't have very much traction outside of Europe or largely affecting the UK is where they get used I don't suppose they get used very much in the states for example then if that matters but it's worth thinking about. I've seen some in bigger clinical trials like multi-site clinical trials not CP specific though.

Conclusion

so just to summarise what we've been hearing, I had the **GOAL** seems to be covering seems to be taking most of the boxes and it needs to be supplemented by objective measurement of walking like **gait analysis**. it may need to be supplemented by **PROMIS** when you're comparing CP to other conditions and maybe some supplemented by objective measure motor functions such as the **GMFM** if in fact it is the focus of interest of the study, and quality of life measure seems to be **CP-QoL** and supplemented again by the **GOAL**.

Adverse Events

One additional domain that we that we will consider is reporting **adverse events** so that is that doesn't necessarily come on the core outcome set but it will be in our recommendation that adverse events of surgery should be reported in a systematic way there's only surgical adverse events measure which we will incorporate this.

Appendix 16 Examples of posters

Development of a core outcome set for lower limb surgical interventions in Ambulant children and young people with cerebral palsy

Hajar Almoajil, Helen Dawes, Sally Hopewell, Francine Toye, Tim Theologies

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BACKGROUND

- Variation in outcome reporting and outcome measures used at varied time point.
- The outcomes of CP trial are not relevant to patients and carer.
- A trend toward selective reporting bias.

AIM

Develop a core outcome set for ambulant children with cerebral palsy and lower limb surgical interventions.

- Minimum set of outcomes and outcome measures
- Studies can be compared and combined
- Informed by key stakeholders

Children and young adults, parents

Healthcare professionals and researchers

METHOD

01 Stage — Identified potential outcomes/outcome measures

- **Qualitative Evidence Synthesis**
  Explore children and parents' experiences of surgery
- **Qualitative Interviews**
  What outcomes are important to key stakeholders?
- **Scoping Review**
  Identified previous reported outcomes
  Identified previous used outcomes measures

RESULT

Wide-range of physical, social and psychological impacts from lower limb surgery on children and families.	
Previous literature confirmed the heterogeneity in outcome domains and outcome measures.	Most published research focuses on outcomes that may not be important to children and parents.
57 outcome domains and 29 outcome measures were identified.	A recent shift toward reporting more patient-centred outcomes.

CLINICAL AND RESEARCH IMPLICATION

- Ensure research focus on outcomes that matter to the children and their parents
- Improve the quality of research, reduce research waste
- Determine core outcome set
- Ensure evidence based clinical practice

02 Stage

- Delphi process
- Consensus meeting

SCAN ME

The need for a core outcome sets for lower limb orthopaedic interventions in ambulant children with cerebral palsy: A Qualitative Evidence Synthesis and Scoping Review

Hajar Almoajil, Helen Dawes, Sally Hopewell, Francine Toye, Tim Theologis

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Introduction

- Lower limb orthopaedic interventions for ambulant children with cerebral palsy (CP) can lead to improvements in motor function

- Evidence for lower limb orthopaedic surgery and post-surgery rehabilitation for cerebral palsy is limited and conflicting

- However, there is a variation in outcome reporting

Purpose

- Synthesise previous reported outcomes in the lower limb orthopaedic interventions for CP literatures

- Determine the potential added value of qualitative synthesis alongside quantitative review of outcomes to inform core outcome set development

Core Outcome Set

- A minimum set of important outcomes

- Studies can be compared and combined

- Determined by multi-stakeholders: Health Professionals , children with CP and families

Methods

- Two separate reviews were conducted:
  - **Qualitative Evidence Synthesis:** Explore children and parents' experiences of lower limb surgery and rehabilitation
  - **Scoping Review:** Identified previous reported outcomes in clinical trials
- Both reviews conducted, using standard methods of:
  - Searching multiple sources
  - Inclusion screening
  - Quality assessment
  - Synthesis
- The outcomes were categorised by using ICF-CY framework
- Thematic synthesis

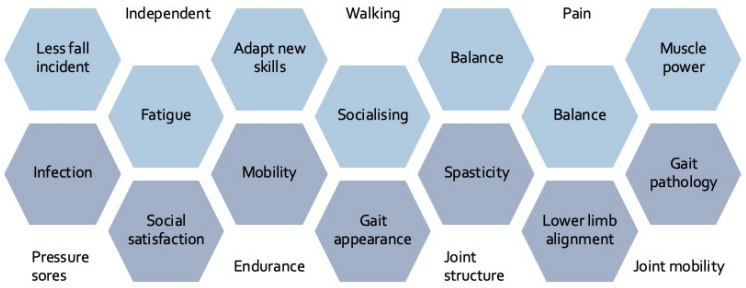
Results

Qualitative Evidence Synthesis

- 6 studies from 4 databases
- 27 different outcomes
- Focus on activity and participation level (e.g., Improve walking ability and to be able to socialise with others

Scoping Review

- 44 studies from 5 databases
- 40 different outcomes reported
- Focus on outcomes related to the child's body function/structure (e.g., lower limb alignment)
- Limited studies reported adverse events of the intervention



Common outcomes identified in Qualitative Evidence Synthesis and Scoping Review

Conclusion

- The reviews highlights a heterogeneity in outcome reporting which undermines the usefulness of research

- Confirmed the need for a core outcome set development to improve the quality of research, reduce research waste

- Qualitative Evidence Synthesis can add value by identifying additional outcomes to those reported in clinical trials and are of importance to children and parents

Implication

- Inform discussions about improving outcome reporting and consistency

- We recommend that the views of all children and their families on the outcomes of importance should be considered when evaluating lower limb orthopaedic interventions

Contact Detail

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References

Health professionals', children' and families' views of the importance of outcomes of lower limb orthopaedic interventions: A qualitative study

Hajar Almoajil, Francine Toye, Helen Dawes, Sally Hopewell, Tim Theologis

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Introduction

- Lower limb orthopaedic interventions for ambulant children with cerebral palsy (CP) can lead to substantial improvements in motor function.

- The reported outcomes do not always take the needs and expectations of children and families into account.

- Qualitative study can enhance our knowledge on perspectives of the different stakeholders of important outcomes.

Core Outcome Set

- A minimum set of important outcomes
- Determined by multi-stakeholders

1. Health professional
2. Children/families

Purpose

Explore the views of different stakeholders on important outcomes of orthopaedic interventions

Methods

- Semi-structured interviews took place in the UK: 10 healthcare professionals, 10 children with CP and 8 parents

- Talking mat: Facilitate conversation with younger children

- Content analysis: International Classification of Functioning-Children and Youth (ICF-CY)
  - ICF linking rule

- Thematic analysis

Results

- Divergence between the outcomes reported by participant group

- A comprehensive list of 31 outcomes were explored

- Categorised into 5 themes:

Body Function and Structure

- Lower limb alignment and symmetry
- Joint flexibility
- Muscle strength
- Pain
- Fatigue
- Mental health

'I would probably measure the general energy levels, fatigue or energy is quite important'

Activity and Participation

- Function in life
- Mobility: sit/stand, transfer and move around, walking and using assistive device
- Participation: within home, school or community
- Social life: leisure activities, sport or hobby

'Feel like hanging out with my mates. But I feel I can't'

Independence

- Daily life activities
- Self care

'I can do many things and independently as well. So, my surgery had a big impact on me being independent nowadays'

Quality of Life

- Emotional wellbeing
- Self-esteem

'The surgery will hopefully help my child have a better quality of life than he would have had without it'

Adverse Events

- Infection
- Fracture
- Wound problem
- Nerve/Blood vessels damage
- Pressure sores

'hopefully avoid any sort of pressure damage at the time'

Conclusion and Implication

- Stakeholders' group described many of the same areas of functioning

- There is notable differences in how these groups prioritise outcomes perspective and construct

- Ensure research focus on outcomes that matter to the children and their parents

- Lead to improvements in clinical practice through relevant outcome measurement

Contribute towards the development of a core outcome set for ambulant children with CP

References and Contact details

The need for a core outcome sets for lower limb orthopaedic interventions in ambulant children with cerebral palsy: A Qualitative Evidence Synthesis and Scoping Review

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INTRODUCTION

- Lower limb orthopaedic interventions for ambulant children with cerebral palsy (CP) can lead to improvements in motor function

- Evidence for lower limb orthopaedic surgery and post-surgery rehabilitation for cerebral palsy is limited and conflicting

- However, there is a variation in outcome reporting

CORE OUTCOME SET

- A minimum set of important outcomes that should be reported in all clinical research

- Studies can be compared and combined

- Informed by key stakeholders: Health professional, children and representatives

PURPOSE

- Synthesise previous reported outcomes in the lower limb orthopaedic interventions for CP literatures

- Determine the potential added value of qualitative synthesis alongside quantitative review of outcomes to inform core outcome set development

METHODS

Qualitative Evidence Synthesis: Explore children and parents' experiences of lower limb surgery and rehabilitation

Scoping Review: Identified previous reported outcomes in clinical trials

- Both reviews conducted, using standard methods of: searching multiple sources, inclusion screening, quality assessment and synthesis

- The outcomes were categorised by using ICF-CY framework

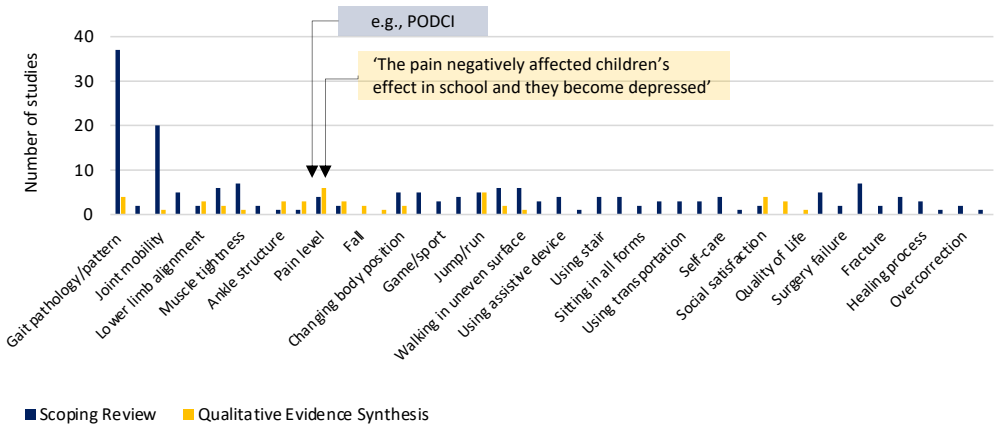
RESULTS

Qualitative Evidence Synthesis

- 6 studies from 4 databases
- 27 different outcomes
- Focus on activity and participation level (e.g., Improve walking ability)

Scoping Review

- 44 studies from 5 databases
- 40 different outcomes reported
- Focus on outcomes related to the child's body function and structure (e.g., lower limb alignment)



CONCLUSION

- The reviews highlight a heterogeneity in outcome reporting which undermines the usefulness of research

- Confirmed the need for a core outcome set development to improve the quality of research, reduce research waste

- Qualitative Evidence Synthesis can add value by identifying additional outcomes to those reported in clinical trials and are of importance to children and parents

IMPLICATION

- Inform discussions about improving outcome reporting and consistency

- We recommend that the views of all children and their families on the outcomes of importance should be considered when evaluating lower limb orthopaedic interventions

REFERENCES & CONTACT DETAILS

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