

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

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## Abstract

**Purpose:** The article identifies the aspects of health and outcomes that are considered important from the perspective of ambulatory children with cerebral palsy (CP) and their parents regarding lower limb orthopaedic surgery and explores how they experience surgical interventions.

**Methods:** Four databases (Embase, MEDLINE (Ovid), CINAHL and PsycINFO) were searched from inception to 11 April 2020. Studies were included if they: 1) they involved children or young adults diagnosed with ambulant CP or their family, 2) participants had experience with lower limb orthopaedic surgery and 3) studies employed qualitative research methods. The Critical Appraisal Skills Programme was used to appraise identified studies. The 'Best-fit framework' synthesis approach was used by applying the International Classification of Functioning-Children and Youth (ICF-CY) linking rules and thematic synthesis. The review process was conducted

according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines.

**Results:** Six studies were included. Four themes were generated which were linked to the ICF-CY framework: Body function and structure, Activity and participation, Environmental factors, Personal factors, as well as non-ICF-CY themes including Emotional well-being and Goal setting. Important surgical outcomes identified were pain, fatigue, movement-related function, mobility, walking ability, community life, emotional well-being, and adequate provision of public and health services.

**Conclusion:** These findings are important for understanding patient-centred outcomes in lower limb orthopaedics surgery and providing focus for future interventional studies aimed at improving outcomes of importance to children with CP. These findings highlight the importance of long-term support to help people negotiate the challenge of surgical regimes and to achieve good outcomes after orthopaedic surgery. The outcomes identified will contribute to the development of a core outcome set in this field.

**Level of evidence:** III

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## Introduction

Cerebral palsy (CP) is the most common physical disability in childhood, currently affecting 2.5 out of 1000 children globally.<sup>1</sup> Its characteristic disabilities, including reduced mobility, persist throughout life and can affect normal development and aging.<sup>2</sup> Mobility in children with CP is generally classified according to the five-level Gross Motor Functional Classification System (GMFCS),<sup>2</sup> with Level I

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indicating good mobility with only slight deficiencies in challenging activities and Level V indicating full immobility and reliance on others.

Children with CP can be defined as ambulatory or non-ambulatory. Ambulatory CP is equivalent to GMFCS levels I–III, and its associated motor impairment is typically managed through physiotherapy followed by orthopaedic surgical procedures.<sup>2,3</sup> Lower limb orthopaedic surgery is undertaken to correct or prevent further deformity and improve motor function.<sup>3</sup>

The World Health Organization International Classification of Functioning, Disability and Health – Children and Youth (ICF–CY) provides a useful framework for assessing people with health conditions such as CP. The ICF–CY incorporates two main parts: (1) body structure and function and (2) activities and participation. In addition, environmental factors (e.g. family and systems) and personal factors (e.g. self-motivation) are considered as barriers or facilitators to these components.<sup>4</sup> The focus of CP management has changed from a concern with the level of impairment to concern with the level of activity and participation as in the ICF–CY framework.<sup>5</sup>

There has also been a noticeable change in perceptions about family involvement in CP management. The approach has gradually shifted from one where health professionals were considered to have all the relevant knowledge,<sup>6,7</sup> to a family/patient-centred care approach, which prioritizes parents and patients over healthcare professionals as being more knowledgeable about patients' needs and abilities.<sup>7,8</sup> However, there is some evidence that the expectations and experiences of children and young people with CP and their parents might differ.<sup>9,10</sup>

To address the issue of unmet expectation and inconsistency of experiences between them, the Core Outcome Measures in Effectiveness Trials (COMET) initiative, developed the concept of a 'core outcome set' (COS) which aims to develop agreed standardized outcome sets for various health conditions and treatments.<sup>11</sup> COS represents the minimum outcome that should be measured and reported in all clinical trials of a specific condition and are suitable for use in both clinical and research settings.

Qualitative Evidence Synthesis (QES) can provide detailed and comprehensive insight into patients' experiences,

perceptions and preferences.<sup>12</sup> It can therefore contribute to the COS development process by providing a preliminary list of outcomes to be considered. Previous QES undertaken to inform COS development in critical type 2 diabetes, bariatric surgery and critical illness have identified important outcomes not reported in systematic reviews of clinical trials.<sup>13–15</sup>

Although previous QES have explored the experience of children and young people with CP,<sup>16</sup> and the experience of their parents in relation to rehabilitation,<sup>17</sup> these QES have not explored the experience of lower limb orthopaedic surgical interventions. There is also no existing QES that aims to contribute to the development of a COS following lower limb orthopaedic surgical interventions for ambulant CP.

The purpose of this QES is to (1) identify the aspect of health and outcomes related to lower limb orthopaedic interventions that are considered important from the perspective of ambulatory children and young people with CP and their parents and (2) identify and synthesize qualitative research that explores how children and young people with CP and their family experience lower limb orthopaedic surgery.

## Materials and methods

The 'Best-Fit' framework approach, which uses a priori themes or models as a starting point, was chosen for the review analysis. Although 'Best Fit' utilizes a deductive approach to data analysis (building on an existing model or framework), it does not ignore data that cannot be accommodated into an a priori framework. These additional data are analysed using inductive thematic analysis (generation of new code emerging from the data).<sup>18</sup> The value of this approach was recognized as combining the strengths of deductive and inductive analysis.

The Best-Fit framework approach follows seven steps as shown in Table 1.<sup>18</sup> The analysis is based on steps 1 to 5 only, as this study does not aim to produce a new framework or conceptual model. This is because, for the purposes of developing COS, it is more useful to provide a descriptive interpretation, rather than the abstracted

**Table 1** Summary of 'Best-Fit' framework synthesis approach

Step	Description
Step 1	Define review question
Step 2	Identify relevant best-fit framework
Step 3	Conduct quality assessment
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

This table is based on Figure 1 from Carroll, C., Booth, A., Leaviss, J. *et al.* "Best fit" framework synthesis: refining the method. *BMC Med Res Methodol* 13, 37 (2013). <https://doi.org/10.1186/1471-2288-13-37>.

conceptual form of QES achieved through steps 6 to 7 'developing a new framework or model'.

The following reporting standards were used: the enhancing transparency in reporting the synthesis of qualitative research statement,<sup>19</sup> supplemented by Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guideline.<sup>20</sup> The review was registered with PROSPERO with the registration ID (CRD42018089538).

### *Step 1: the review question*

The SPIDER approach (Sample, Phenomenon of Interest, Design, Evaluation, and Research type) was used to define the research question.<sup>21</sup> The SPIDER question was formulated as follows: What are the key health outcomes following lower limb orthopaedic surgery from the perspective of ambulant children and young people with CP and their parents?

### *Step 2: search and study selection*

#### *Identification of a relevant framework*

The ICF–CY was selected as the 'Best-Fit' framework. It was chosen for two reasons: (1) the generated themes would represent a robust base of knowledge that form the first list of a future COS for lower limb orthopaedic surgery of ambulant children and young people with CP; and (2) the ICF–CY terminology is considered a global framework and common language within CP studies, therefore, results can be linked to previous studies in the field of CP.

#### *Identification and selection of relevant primary studies*

A search was performed using four databases (Embase, MEDLINE (Ovid), CINAHL and PsycINFO) from inception to 11 April 2020. Supplementary File 1 lists the keywords used in the search and their combinations from each database. A Web of Science citation search and a review of references from each included study to identify any additional studies was conducted.

The titles and abstracts of studies identified were screened for the following inclusion criteria: (1) the study population consisted of individuals diagnosed with CP and/or their parent or primary caregiver; (2) the individuals were considered ambulatory or within level I, II or III of the GMFCS; (3) the CP individuals had undergone lower limb orthopaedic surgery; (4) the study employed qualitative research methods; (5) in the case of mixed-method studies, data from the quantitative and qualitative approaches were reported separately; and (6) the full article was published in English.

Opinions, editorials and studies containing only quantitative data were excluded. Studies with samples representing several types of conditions were excluded if the authors had not defined findings by type or specified

which data was linked to individuals with CP. In addition, studies only reported as an abstract or published as a poster were excluded, as it was not possible to adequately assess their methodological quality.

To ensure the transparency of the process and to deal with uncertainties,<sup>22</sup> all members of the study team had a role in screening and identifying studies.

### *Step 3: quality appraisal and data extraction*

#### *Quality appraisal*

All the included studies were assessed using two methods of appraisal: (1) the Critical Appraisal Skills Programme (CASP),<sup>23</sup> and (2) the global categorization, described by Dixon-Woods et al: 'key paper' (conceptually rich and could potentially make an important contribution to the synthesis), 'satisfactory paper', 'uncertain' (which needs to be screened by an additional reviewer), 'irrelevant' to the synthesis, and a methodologically 'fatally flawed'.<sup>24</sup>

The CASP appraisal was not used as a basis to exclude studies from the review, but rather, it allowed the reviewers to consider the merits of each paper. In contrast, studies classified as irrelevant or fatally flawed were excluded. Those categorized as key and satisfactory papers were included in the review.

Two reviewers independently appraised all relevant studies identified during CASP assessment (HA, JPa) and on the basis of the global categorization described by Dixon-Woods and colleagues (HA, FT).

#### *Data extraction*

A template was created for extracting data from the identified studies. The major elements included: study setting, population, participant characteristics, the intervention delivered, methodological design and approach taken by the study; methods for identifying the sample recruitment; and the data collection and analysis methods.

### *Step 4: coding evidence against a priori framework*

In order to link captured information to precise ICF–CY codes, the ICF–CY linking rules were used.<sup>25</sup> The rules advise utilization of ten linking rules as shown in Table 2. Each line of text was separately coded according to its meaning. Each code in the data linked to the most precise ICF–CY code.

If the content of a code was not explicitly named in the ICF–CY category, the 'other specified', 'unspecified', 'not defined', 'not covered' and 'health condition' category was applied, as shown in rules 3 to 5 and 7, respectively. In order to assess the reliability of coding, two reviewers (HA, JPi) independently linked the data to the ICF–CY taxonomy.

**Table 2** 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 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 on within a certain piece of information when linking it to the ICF.
5	Identify and document the categorization 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 nd (not definable).
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 pf (personal factors).
10	If the meaningful concept is not contained in the ICF, assign this meaningful concept to nc (not covered).

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### Step 5: creating new themes

A thematic analysis defined by Thomas and colleagues<sup>26</sup> was conducted where the data did not precisely fit the framework. New categories were created to capture the essence of meaning of the initial codes. The categories were discussed and refined by three reviewers (HA, FT and JPi) to develop a final list of relevant themes that explained experience and outcomes from the stakeholders' perspective.

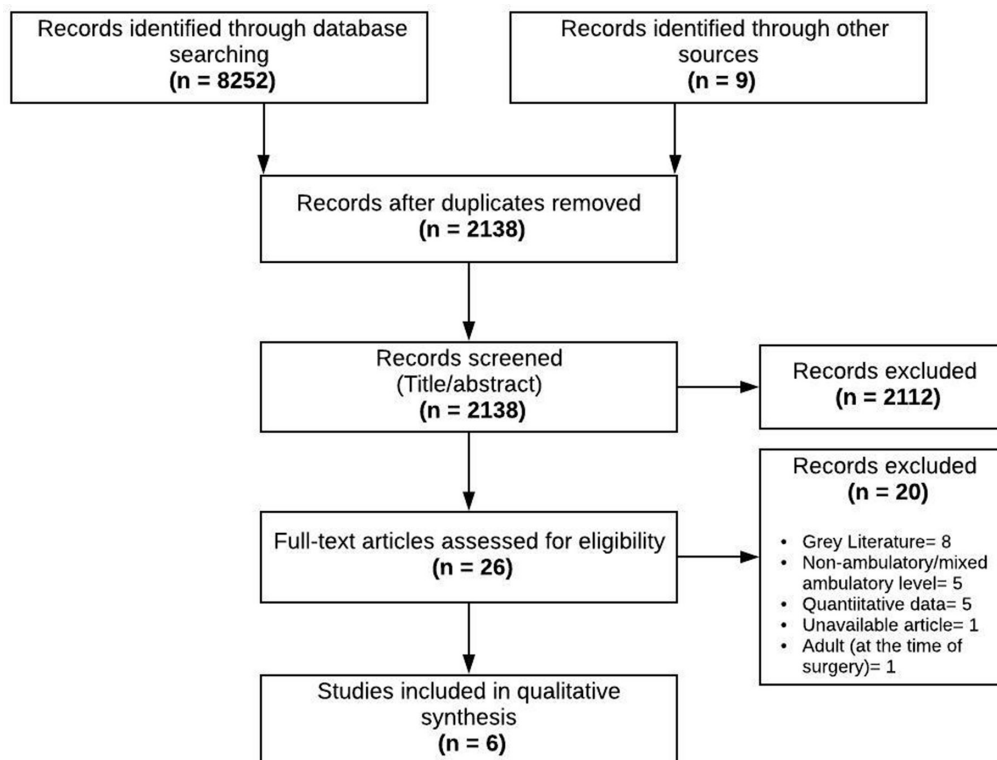
### Results

The literature search initially identified 8252 studies. A manual search of the references cited in the available reviews of CP yielded an additional nine abstracts. All identified studies were imported into an EndNote library (Endnote X8), duplicates were removed (6123) and the remaining 2138 title/abstracts were screened, with 26 being selected for full-text review. Of these, 20 were excluded: five did not provide qualitative data, five did not

**Table 3** Characteristics of included studies

Authors	Country	Participants	Age	GMFCS	Surgery	Time from surgery	Interview methods	Analysis	Study aim
Capjon et al, 2010 <sup>27</sup>	Norway	8 children and parents	12–16 years	–	SEMLS	6 and 12 months	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 et al, 2010 <sup>28</sup>	Norway	8 children and parents	12–17 years	I, III, III	SEMLS	Prior to surgery	Semi-structured interview	Kvale's method	Explores the pre-operative situation of children accepted for multilevel surgery for cerebral palsy and their parents
Iversen et al, 2009 <sup>31</sup>	Norway	12 parents	8–16 year	–	Not specified	Post-surgery hospitalization	Semi-structured interview	Phenomenal	Explores parents' experiences when their child who is disabled with cerebral palsy was going through a surgical procedure
Lehtonen et al, 2015 <sup>30</sup>	Finland	10 young adults	15–22 years	II, III	SEMLS	5 years post-surgery	Semi-structured interview	Phenomenal	Examine 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
Høiness et al, 2014 <sup>29</sup>	Norway	7 children and parents	9–16 years	III	SEMLS	1 and 5 years post-surgery	Semi-structured interview	Kvale's method	Investigated children who underwent SEMLS regarding post-operative rehabilitation and pain, gait parameter 1 year after surgery and mobility 5 years after surgery
Stephan-Carlier et al, 2014 <sup>32</sup>	France	11 children and parents	13–21 years	I, II, III	SEMLS	At least 1-year post-surgery	Semi-structured interview	Descriptive	Confront the perceptions of parents and the experience of their operated children in terms of quality of life and surgical outcome

Note. (–) reported 'ambulatory' to describe mobility level of the participants  
SEMLS, Single Event Multilevel Surgery



**Fig. 1** Flowchart of the studies selection process.

provide data of ambulatory CP, eight were grey literature (conference abstract), one was based on a perspective of adult CP population, and one study's data was unavailable (Supplementary File 2). The synthesis included six studies on the experience and perspective of children and young people with CP and their parents on lower limb orthopaedic surgery (Table 3).<sup>27–32</sup> The selection process details are outlined in Fig. 1.

CASP appraisal for included studies for aspects such as recruitment, data collection methodology, data analysis and relevance of the findings were either high or moderate. However, often studies did not describe the 'relationship between researchers and participants' and 'ethical issues'.

#### Synthesis finding

The analysis created a total of 26 initial codes related to the ICF–CY framework. These codes were categorized into ten and four ICF–CY chapters and themes, respectively. The resulting themes include: (1) Body function and structure, (2) Activity and participation, (3) Environmental factors and (4) Personal factors. Additionally, four codes that did not fit the ICF created two additional themes: (1) Emotional well-being and (2) Goal setting. Distribution of the studies supporting each ICF–CY concept and each of the two additional themes are available in Supplementary File 3). A list of the identified outcomes summarized in Table 4.

#### Theme one: Body function and structure

All six studies reported treatment experiences related to the 'body function and structures' components of the ICF–CY framework. Of eight chapters found under 'body function and structure', the data linked to the following three, functions of the cardiovascular and respiratory systems (Fatigue), sensory function and pain, and neuromusculoskeletal and movement-related function and structure.

##### *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 their child keeping up with friends:

*'He has to stay in the classroom during breaks, it costs [energy] too much to go out.'*<sup>28</sup>(Parent)

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

##### *Sensory function and pain*

Reduced pain was also described as an important outcome post-surgery. Some children reported less pain



**Table 4** Desired outcomes

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

compared to pre-surgery while others, along with their parents, defined unanticipated pain post-surgery that negatively impacted upon their daily life, such as limiting inclusion in community and school activities and causing sleepless nights and leading to 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.'*<sup>27</sup>(Parent)  
*'I tried to do the same as the other kids, got knee pains, and ended in a wheelchair for a week.'*<sup>28</sup>(Child)

Participants described a range of potential sources of pain post-surgery, including the application of a cast post-surgery and limited support or adjustment of facilities at school or home, making mobility more painful. This pain could exacerbate 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.'*<sup>27</sup>(Parent)

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

#### *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. This included improvement in joint range of motion, muscle power and muscle endurance. Parents and children felt that these improvements would lead to restored balance, reduced 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.'*<sup>30</sup>(Child)  
*'We're worried about her back, knees and hips. We hope she'll have fewer [Neuromusculoskeletal] symptoms in the long run.'*<sup>28</sup>(Parent)

However, 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 treatment experience 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, including Mobility and Community, social and civic life.

### 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, faster and for long distances. Studies indicated that developing and acquiring new mobility skills, such as running and moving around, was another important surgical outcome of children and their parents.

*'Maybe I'll be able to walk a bit more like the others. Maybe.'*<sup>27</sup>(Child)

*'... 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.'*<sup>27</sup>(Child)

*'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.'*<sup>27</sup>(Parent)

However, some studies were less positive, for example, there were reports of children losing their ability to walk or eventually being in 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.'*<sup>27</sup>(Child)

### Community, social and civic life

Studies indicated that children, young people and their parents described improved interactions and social participation as an important outcome. The experience of participation was reported at three levels: school, community and family activity.

*'Now I hang around more with the other boys in the class; I couldn't do that before.'*<sup>27</sup>(Child)

*'We have achieved the goals we set for him, ..., manages to keep up with the others and has participated in gym during the past six months. .... We are very pleased.'*<sup>27</sup>(Parent)

Being socially accepted was another important goal of undertaking surgery for both children.

*'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.'*<sup>28</sup>(Child)

### Theme three: Environmental factors

Four studies reported 'environmental factors' that enhanced or inhibited positive outcomes. Of the five chapters found in 'environmental factors' in the ICF-CY framework, the data linked to three chapters; Support and relationships, Attitudes, and Services, systems and policies.

#### Support and relationships

Health professionals' support was considered to play an important role in the success of treatment and goal achievement. Parents viewed health professionals' support positively and described it to be 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.'*<sup>27</sup>(Parent)

Studies reported the importance of support for the child undergoing surgery. Children referred to support as positive interactions during the clinical sessions, and also encouragement from families, friends, peers and health professionals.

*'A lot of responsibility was loaded on me. I need understanding and support.'*<sup>30</sup>(Child)

*'The lady in the cafeteria helps me out.'*<sup>30</sup>(Child)

In contrast, several parents reported concerns regarding lack of, or insufficient, information. Doctors were criticized for not listening and for talking in medical terminology that families could not fully understand. These concerns impacted upon the parents negatively in term of feeling uncertain and ambivalent about their desired outcome post-surgery, and also feeling incompetent and powerless. This limited their ability to make appropriate decisions regarding the treatment. Consequently, they sought information and support from other families that 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.'*<sup>31</sup>(Parent)

*'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.'*<sup>28</sup>(Parent)

*'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.'*<sup>27</sup>(Parent)

### Attitude

Parents described negative attitudes from school peers in the form of physical and verbal bullying as an experience that could have a negative effect on a child's well-being. As the extreme, surgery could be seen as a way to move themselves away 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.'*<sup>28</sup> (Parent)

### Services, systems and policies

The quality and availability of the healthcare system were factors that either facilitated or inhibited good outcome from surgery. Parents frequently indicated that follow-up waiting times, the setting of interventions (i.e. home, school or community), the lack of highly qualified health professionals and the presence of inadequate facilities, such as lack of wheelchair access, acted as barriers that negatively affected their child's potential to achieve treatment outcomes.

*'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 6 months without the prospect of any support other than that of our capable but inexperienced physiotherapist.'*<sup>27</sup> (Parent)

### Theme four: Personal factors

Six studies reported personal motivation as a 'personal factor' that might have an effect on achieving a good treatment outcome.

Studies described being motivated by future goals such as improved activities of daily life and being able to socialize with other children. In contrast, post-surgery complications, unmet needs and negative previous surgery experiences 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.'*<sup>27</sup> (Child)  
*'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.'*<sup>27</sup> (Parent)

### Theme five: Parents' emotion and well-being

One study reported the impact of surgery on parents' 'emotion and well-being'. A strong sense of responsibility

and dedication to their children had a profound impact on families who could feel vulnerable and frustrated:

*'Sometimes unpredictable tasks every day, and a hospitalization gives even more tasks, worry and tiredness.'*<sup>31</sup> (Parent)  
*'I experienced a lot of stress, but you have to be strong.'*<sup>31</sup> (Parent)

### Theme six: Goal setting

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

*'The physiotherapist knows best how I should train.'*<sup>27</sup> (Child)  
*'She (referring to her physical therapist) lets me decide a lot, it feels really good.'*<sup>30</sup> (Child)

## Discussion

This QES aimed to provide an overview of important outcomes and experiences that might affect these outcomes among ambulant children and young people with CP and their parents, in relation to a lower limb orthopaedic surgery. Through the best-fit synthesis of six studies, a number of outcomes of importance to the stakeholders were identified and categorized under the guidance of the ICF-CY framework: pain management, improvement in the symptoms and clinical characteristics of the condition (joint mobility, muscle power, balance, gait appearance and its quality, walking and being able to socialize with others). A number of experiences, including the impact of the surgery and stress of rehabilitation, the importance of communication and support of health professionals, were highlighted by both parents and young people. It also highlighted the emotional impact on parents. The findings suggest a need for a range of measures to capture these important outcomes. There is also a need for clear information and support both in preparation and during the rehabilitation process following surgery.

These findings show that pain, fatigue and gait abnormality impact on the ability of the children undergoing surgery to function socially. This may reflect that the symptoms of CP commonly manifest as limitations of physical function, pain and fatigue as a result of joint contracture, bone deformities and muscle weakness. These, in turn, can lead to a deterioration in gait and function over time in ambulant children.<sup>33</sup> The QES highlights 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 as part of the 'body function and structure' impacted on the persons' ability to



socialize with other children. This finding is in agreement with another review of the patient perspective on rehabilitation services, which found that pain and fatigue restricted children and young people with CP daily life activity.<sup>16</sup>

There are contrasting assumptions on whether activity and participation represent important post-surgical outcome for CP. It has been 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'.<sup>34</sup> However, Narayanan (2016) suggested that lower limb orthopaedic surgery for ambulatory children with disability should focus on improving the quality of gait (i.e. more symmetry, reduced reliance on walking aids) and on optimizing gait efficiency through pain prevention, preservation or improvement in physical function, activity and participation.<sup>35</sup> Therefore, involvement of key stakeholders, including individuals with CP and their parents, alongside health professionals during selection of appropriate outcomes for lower limb surgical trials is recommended to enhance understanding of the patients' preferences and values.

Environmental and personal factors were shown to shape the experience of lower limb orthopaedic surgery. For instance, lack of appropriate equipment and local services to support post-surgery rehabilitation were highlighted as being a barrier to the positive effects of the lower limb surgery. Participants identified the importance of support and communication with health professionals. Effective communication between patients, family and health professionals is paramount in optimizing the partnership, which in turn underpins the positive outcomes of the surgery. However, this review and other studies conducted within the CP population in different healthcare settings concluded that a paternalistic approach might still be prevalent.<sup>16,17,36</sup> Hence, input from the children and young people and families might enhance the interaction with the health professionals.

While the concept of 'personal factors' is not well defined within the ICF-CY framework, the World Health Organisation (WHO) emphasizes the importance of personal factors in the intervention's outcomes.<sup>4</sup> When reviewing the personal factors that may affect experiences related to the surgery, a list of demotivating factors was identified. This included previous symptoms, past experiences from surgery, prolonged rehabilitation before the benefits of surgery were evident and the significant effort required during the rehabilitation process. Although lack of motivation was recognized as likely to have an impact on treatment adherence and outcomes, this lack of motivation could also be attributed to damage to the motivation control areas in the brain due to the condition.<sup>37,38</sup>

Studies have sought to determine the impact of age, sex and GMFCS levels on parental satisfaction, children's

motivations and ability to cope with the post-operative regime, and health outcomes such as pain and functional mobility.<sup>39-43</sup> For example, Svehlik and colleagues (2011) highlight that a growth spurt at puberty might influence Single-Event Multilevel Surgery (SEMLS) outcomes as this growth spurt contributes to the development of muscle contractures,<sup>44</sup> and therefore might be responsible for increased pain.<sup>45</sup> These factors did not influence the children's or parents' experiences in this synthesis. This may be due to the small number of studies included. Future qualitative research exploring the experience of particular sub-groups might help us to understand whether or not these factors affect experiences and priorities.

A family-centred care approach emphasizes the importance of collaboration between families and health professional in goal setting and decision making.<sup>46</sup> This approach has been shown to lead to more effective outcomes and to be more efficient in terms of the intervention required by professionals.<sup>47</sup> However, the extent to which the process involves the child with CP and their family is not clear. In the current QES, children and young people and their family's perspective towards goal setting was varied. This variation was supported by another mixed studies review investigating the experiences of parents of children with CP in rehabilitation services.<sup>17</sup>

Finally, parents report overwhelming levels of stress and vulnerability, which tends to be particularly associated with post-surgery hospitalization, the rehabilitation process and insufficient information and resources. This description of the impact of their child's surgery on the parents resonates with a previous study on the well-being of parents of children with CP. This found that the parents' self-perception, family functioning and psychological well-being are affected by the child's disability.<sup>48</sup> This highlights the need for further parental psychological support.

This synthesis has drawn a broad overview of experiences at different time points following surgery, and it may be that there is a relationship between children's and parents' concerns and the time point post-surgery. For example, qualitative studies that report experiences in relation to post-surgical pain within a short time period of surgery might have a different emphasis to those that recruit later. Studies indicate that children and parents report persistent post-surgical pain at 12 months after major surgery,<sup>29,49,50</sup> which is associated with functional disability and decreased quality of life. In another example, there may have been a greater exploration of and emphasis on parental emotions and well-being had one study recruited parents from the post-operative hospitalization period. Again, the fact that it was close to the surgery, within just a few days of post-operative experiences, might have affected how the parents feel about the whole experience,<sup>51</sup> therefore, interviews at this time might place an emphasis on emotional impact. Future qualitative

research drawing on experiences from different points in time would contribute to our understanding of important outcomes for young people and their families.

This QES had several important strengths. To our knowledge, this is the first QES using best-fit synthesis in the CP orthopaedic surgery literature for outcome prioritization aiming at COS development. The review was conducted by at least two reviewers at all stages, including (1) the study selection, (2) the data extraction and (3) the synthesis process.

This review has its limitations. Six studies were included in this review and the majority were from one country. Healthcare services may differ among countries. Most of the studies' participants had undergone SEMLS and there may be different experiences and concerns from children who have one or two procedures at any one time. There was a lack of directly relevant evidence; that is, no study was conducted for the explicit purpose of determining children and young people's or parents' priorities for orthopaedic treatment outcomes. In addition, the inclusion of all studies, regardless of their quality, may have influenced the findings: some of the included studies had limited qualitative data to extract which may have influenced our interpretation. Future in-depth qualitative studies exploring the experience of surgery would contribute to our understanding of children with CP and their parents and therefore contribute to improvements in healthcare.

In conclusion, this review identified key outcomes that should be assessed in the clinical and research settings as they are important to children and young people with CP and their parents. The identified outcomes should be considered in the development of a core outcome set for lower limb orthopaedic surgery for ambulant CP. There is a limited number of qualitative studies identified in this field ( $n = 6$ ) and it is important to incorporate patient perspectives from the outset of COS development. Future qualitative research should focus on lower limb orthopaedic surgery outcomes in ambulant CP children and their families.

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## COMPLIANCE WITH ETHICAL STANDARDS

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## ETHICAL STATEMENT

**Ethical approval:** This article does not contain any studies with human participants performed by any of the authors. Ethical approval was not required for this work.

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## ICMJE CONFLICT OF INTEREST STATEMENT

None declared.

## AUTHOR CONTRIBUTIONS

HA: Designed the review protocol and helped to draft the manuscript, Completed the systematic review search and extraction of eligible papers, Contributed in quality appraisal of the included studies, Analysed the data, Wrote the first draft the manuscript, Edited the manuscript and read and approved the final version.

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SH: Designed the review protocol and helped to draft the manuscript, Edited the manuscript and read and approved the final version.

FT: Designed the review protocol and helped to draft the manuscript, Completed the systematic review search and extraction of eligible papers, Contributed in quality appraisal of the included studies, Analysed the data, Edited the manuscript and read and approved the final version.

## SUPPLEMENTAL MATERIAL

**Supplemental material** is available for this paper at <https://online.boneandjoint.org.uk/doi/suppl/10.1302/1863-2548.14.200139>

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