A global core outcome set for orthopaedic interventions in children with spinal dysraphism

aiming to enhance research quality and patient outcomes

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Aims

Orthopaedic interventions in spinal dysraphism (SD) are frequently required to address a wide spectrum of musculoskeletal deformities. The outcomes used to assess treatment, however, are heterogeneous and most fail to incorporate patient/family perceptions. The aim of this study was to identify the minimum set of outcomes to be collected in clinical practice and research settings following orthopaedic intervention in ambulatory and non-ambulatory children with SD.

Methods

The study was based on Core Outcome Measures in Effectiveness trials (COMET) initiative. A list of individual clinical outcomes (ICOs) and outcome measurement tools (OMTs) were obtained from a systematic literature review (SR) and from patients and families through an interview and questionnaire. Core outcomes were rated for importance in a two-round Delphi process that included international orthopaedic surgeons, physiotherapists, orthotists, patients, and families. Outcomes that did not reach consensus during the Delphi process were resolved with a final consensus meeting.

Results

In total, 88 statements, including ICOs and OMTs, were scored during the Delphi process for ambulatory and non-ambulatory children. A total of 35 items were resolved in the final consensus meeting. The final core outcome set (COS) is goal-based and includes 28 outcome parameters to be collected a minimum of one year after any orthopaedic intervention and at subsequent set points during childhood. The COS incorporates clinical examination, mobility and functional assessment, patient-reported outcome measures, and investigations with the Goal Attainment Score recommended for goal setting.

Bone & Joint Open

Conclusion

A minimum set of outcomes to evaluate the orthopaedic treatment of SD was created thereby enabling consistency in reporting among centres and studies.

Take home message

- A minimum set of outcomes to evaluate the orthopaedic treatment of spinal dysraphism was created enabling consistency in reporting among centres and studies.
- The core outcome set incorporates clinical examination, mobility and functional assessment, patient-reported outccome measures, and investigations with the Goal Attainment Score recommended for goal setting.

Introduction

Spinal dysraphism (SD) refers to a range of congenital open and closed neural tube defects with a multifactorial aetiology resulting in a spectrum of neurological abnormalities.[1] Patients with SD develop impairments and disabilities arising from the spinal cord dysfunction, as well as associated abnormalities of the brain. Orthopaedic interventions such as surgery, orthotic use, and pressure area care are used to address a wide spectrum of musculoskeletal issues.[2,3] Orthopaedic interventions are needed in almost every child with SD with the goal of optimizing function and role participation now and for the future.

The outcomes used to evaluate orthopaedic interventions reported in the literature are heterogeneous and seldom incorporate patient or family perceptions.[4] Variability in care is exacerbated by the lack of standardization in outcome reporting.[4]

The Core Outcome Measures in Effectiveness trials (COMET) initiative has established guidelines to develop core outcomes sets (COSs),[5] a minimum standardized set of outcomes to be reported in all studies investigating a specific clinical condition. The use of COSs is well-established in paediatric orthopaedic clinical research and practice.[6-10] Standardization of outcomes reporting for studies investigating treatment options for SD will allow meaningful meta-analyses to be made and will lead to clear guidance for the orthopaedic management of this condition.

This study aims to identify a minimum set of outcomes to be collected in clinical practice and research following orthopaedic intervention in children with SD according to the COMET guidelines.

Methods

The study followed the protocol for development of a COS for the orthopaedic management of SD which has been previously published.[11] The process included four stages:

1. Identification of outcomes reported in the literature through a systematic review;[4]
2. Identification of outcomes important to patients and carers through interviews and questionnaires;
3. Scoring the list of outcomes obtained from the previous stages, as well as adding any novel outcomes, through an international Delphi process; and
4. A final consensus meeting.

For the purposes of this study, the term 'orthopaedic intervention' refers to both non-surgical and surgical treatments for musculoskeletal conditions, ranging from noninvasive methods such as casting, orthotic use, and skin/wound care to surgical procedures that often concentrate on deformity correction.

Systematic review

An initial list of outcomes reported in the literature was identified through a systematic review, and has been published.[4]

Questionnaires and qualitative interviews

Qualitative semi-structured interviews led by the treating clinician were held with carers and children during routine outpatient visits according to the study protocol.[11] The interviews identified the key outcomes of SD among families using study-specific questionnaires designed for both patients and carers. A purposive sampling strategy, including 32 patients and carers,[12] was used to identify outcomes normally not considered in clinical and research settings. The questionnaires for the carers were completed as a self-reported questionnaire complemented by a semi-structured interview with the researcher, who used the carers' answers as a prompt for further discussion. The questionnaires for the children were grouped according to age and were completed by the patients or with carers' help when needed. This is a well-established method used commonly in similar projects.[13,14]

Ethical approval

This project was prospectively registered (AUDI004001). On consultation with the St George's NHS Healthcare Trust-research and development department, this project was deemed a service evaluation. No patient identifiable information was collected.

The Delphi survey

The outcomes identified in the first two stages served as the foundation for an international Delphi process,[15] which included two rounds of voting, each lasting six weeks. The outcomes identified in the systematic review were discussed, refined, and structured during a steering meeting before the first round. In the first round, participants' demographic information, including contact email, stakeholder group, and country, was collected. The participants were then asked to score the list of suggested outcomes from 1 to 9 (with 1 to 3 being not relevant, 4 to 6 being important but not critical, and 7 to 9 being extremely relevant). The list of outcomes divided into those applicable to 'ambulatory' and 'non-ambulatory' patients included outcome measurement tools (OMTs), individual clinical outcomes (ICOs) and investigations (Ixs). Participants were given the opportunity to suggest additional outcomes of relevance not already listed and were asked for their ideas on the minimum postintervention follow-up and the relevant timepoints for COS collection to ensure data

Table I. Delphi survey participants groups.

Round	Respondents, n (%)
Delphi round 1	**42**
Consultant orthopaedic surgeon	28 (67)
Orthotist	2 (5)
Physiotherapist	5 (12)
Parent/carer	7 (17)
Delphi survey round 2	**39**
Consultant orthopaedic surgeon	26 (67)
Orthotist	2 (5)
Physiotherapist	4 (10)
Parent/carer	7 (18)

were meaningful for both ambulatory and non-ambulatory children.

To maximize engagement with lay participants (e.g. children, parents, and families), the initial section avoided medical terminology and technical details. All participants were encouraged to complete this section, but only healthcare professionals were asked to address the subsequent section focused on technical outcomes of success.

The data obtained from round one were analyzed using bar charts stratified by 'ambulatory' and 'non-ambulatory' groups. The data were summarized as 'consensus in', 'consensus out', or 'no consensus' based on the Grading of Recommendations, Assessment, Development and Evaluations (GRADE) guidelines,[16] where:

1. Consensus in was defined as > 75% of the group rating the outcome extremely relevant (7 to 9 points), with < 15% rating the outcome not relevant (1 to 3 points).
2. Consensus out was defined as > 75% of the group rating the outcome not relevant (1 to 3 points), with < 15% rating the outcome extremely relevant (7 to 9 points).

Outcomes that reached 'consensus in' were added to the final core outcome list. Outcomes that reached 'consensus out' were removed from the list and outcomes that reached 'no consensus' were added to round two of the Delphi survey, along with any additional outcomes suggested during round one. During round two, participants were invited to again score the outcomes that did not reach 'consensus in' during round one using the same descriptors and with the option of changing their scores if they chose to. Data obtained from round two were then summarized using the GRADE guidelines as in round one.

Following round two, outcomes that reached 'consensus in' were added to the final core outcome list, outcomes that reached 'consensus out' were removed, and outcomes that reached 'no consensus' were added to the final consensus meeting stage.

Final consensus meeting

The list of outcomes obtained from the Delphi survey was presented in a final consensus meeting which was held in May 2024. The hybrid meeting was attended by 12 international stakeholder representatives, including orthopaedic surgeons, physiotherapists, nurse practitioners, and parents/patients' representatives. First, the full list of outcomes included in the Delphi survey was presented, with outcomes divided according to whether they reached 'consensus in', 'consensus out', or 'no consensus'. There was an opportunity for an open discussion related to all outcomes as well as minor rephrasing and the amalgamation of outcomes when needed. Outcomes that did not reach consensus were discussed and voted upon with an 'inclusion yes/inclusion no' procedure, where the score of the majority (51% of the voting participants) decided the outcomes: inclusion or exclusion from the final set. Participants scored each outcome simultaneously on their computer screen. The scores were counted and reported by an independent member.

Following the final consensus meeting

The final list of outcomes generated through the entire process was compiled and structured into the COS assessment form. The final draft was approved via email by the stakeholder representatives who participated in the consensus meeting.

Results

The participant distribution is presented in Table I.

The list of collected outcomes, the stage it was identified in, and the decision following the Delphi process is presented in Table II for the ambulatory child, Table III for the non-ambulatory child, and Table IV for the investigations and outcome measurement tools (OMTs). A flowchart of the number of outcomes collected and processed at every stage is presented in Figure 1.

Systematic review

The outcomes collected in the systematic review were published in 2024.[4] In summary, after data extraction, 27 individual clinical outcomes (ICOs) (of which four were Ixs and ten outcome measurement tools (OMTs)) were identified and categorized in domains according to the Outcome Measures in Rheumatoid Arthritis Clinical Trials (OMERACT) modified filter.[40]

The 23 non-investigation-based ICOs were categorized into: mobility (n = 4); clinical assessment (n = 12); adverse events (n = 6); and miscellaneous (n = 1).

During the steering group meeting held before the Delphi survey, the 23 ICOs from the systematic review were expanded into 30 statements. Of these, 21 were applicable to both ambulatory and non-ambulatory individuals, five were specific to ambulatory individuals, and four were relevant only to non-ambulatory individuals. This created a total of 51 ICOs (26 ambulatory and 25 non-ambulatory).

Of the four Ixs from the systematic review, two remained unchanged, one was removed after discussion, one was added, and one was expanded into six Ixs (a general radiology statement expanded into specific radiological parameters for different joints) which resulted in nine Ixs in total.

The 51 ICOs, ten OMTs, and nine Ixs resulted in a total of 70 statements.

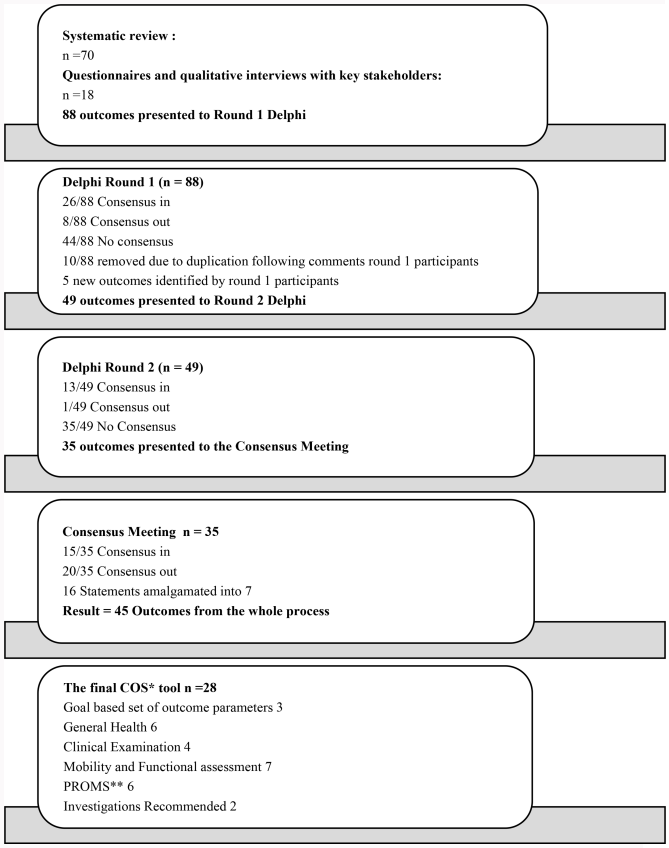

Fig. 1
Flowchart of the core outcome set process, and the outcomes identified at each stage. COS, core outcome set. PROMs, patient-reported outcome measures.

Questionnaires and qualitative interviews

In all, 13 patients and 19 carers, under the care of a tertiary multidisciplinary SD clinic participated in the interviews and questionnaires process as a part of their appointment at St George's Hospital and Great Ormond Street Hospital, UK.

Two OMTs were included as well as nine ICOs specific to ambulatory and seven to non-ambulatory patients. This resulted in a total of 18 items (two OMTS and 16 ICOs) added to the list (Tables I and II). A total of 88 statements were included in Delphi round 1.

It is important to highlight that, for both ambulatory and non-ambulatory children, all outcomes classified under the 'quality of life: wellbeing, activity, and participation' domain including emotional wellbeing, school attendance, participation in sports, and social engagement with peers were incorporated during the interviews and questionnaire phase of outcome collection.

Delphi survey

Round one: There were 42 respondents to the first round of the Delphi survey from 12 countries. The stakeholders were categorized into two groups: healthcare professionals (83%) and individuals with SD and their carers (17%). The participant distribution is presented in Table I. The respondents were from the UK, Turkey, Egypt, Germany, the Netherlands, Portugal, USA, Canada, Brazil, Italy, Austria, and India.

The list of statements presented to round one of the Delphi was comprised from the outcomes collected from the SR (n = 70), together with the 18 outcomes identified during the patients/parents' questionnaire/interview, providing a total of 88 outcomes (Figure 1).

Of the 88 items included in round one, 26 were 'consensus in', eight were 'consensus out', 44 reached 'no consensus', and ten items (nine ICOs and one Ix) were removed following comments from round one participants. Multiple stakeholders stated that there was repetition and duplication of statements, reporting the same outcome in a different way which was confounding the process. These are summarized in Appendix 1 of the Supplementary Material.

Five additional OMTs were identified by round one participants and added to the list for presentation to round two (Figure 1, Tables II and III).

The descriptive analysis of the stakeholders' responses to outcomes in relation to the ambulatory and non-ambulatory child Delphi survey round one is available in Appendix 1 of the Supplementary Material.

Round two: The number of respondents to round two was 39 (7% drop out rate), which was made up of 82% healthcare professionals and 18% patients/parents. The participant distribution is presented in Table I.

Of the total 49 items listed for scoring in round two, 13 obtained 'consensus in', one obtained 'consensus out', and 35 obtained 'no consensus' and were put forward to the consensus meeting. The minimum time for follow-up before COS collection was determined as one year after any intervention with three set collection points during childhood: starting school, end of primary school, and skeletal maturity.

The descriptive analysis of the participants' responses to importance of outcomes in relation to the ambulatory and non-ambulatory child Delphi survey round two is available in Appendix 2 of the Supplementary Material.

Final consensus meeting

The meeting was a hybrid meeting including 12 participants with six attending face to face and six attending virtually. The meeting participants were from the UK, India, Brazil, Italy, Canada, and USA. It included eight orthopaedic surgeons, two advanced practitioners, and two parent/patient representatives. The voting was coordinated by an independent facilitator (JM). The parent representatives participated only in the voting procedure related to non-technical outcomes, which included areas of adverse events, life impact, and pathophysiological manifestations.

Of the total 35 items voted upon in the meeting, 15 were voted as 'consensus in' and 20 'consensus out'. Only one OMT reached 'consensus in' during the Delphi process (goal attainment scale in rehabilitation (GAS)).[17,41]

The number of outcomes reaching 'consensus in' from round one was 26, from round two was 13, and with 15 from the consensus meeeting, resuted in a total of 54 outcomes. 16 statements were amalgamated into seven for clarity and ease of use (for example statements 8, 10, 12, 17, 37, and 38 were combined into one statement applicable for both ambulatory and non-ambulatory: 'no foot or ankle deformity that interferes with splint use, footwear and/or function including standing frame use') resulting in 45 statements.

The total number of outcomes collected from the entire Delphi process is 45 (41 ICOs, three Ixs, and one OMT). The items that were voted upon and their results

Table II. The ambulatory child: the collected individual clinical outcomes, stage of collection, and voting.

Individual clinical outcomes (mapped to ICF domains)	Stage of collection	Delphi stage 1 vote	Change to statement stage 2	Delphi stage 2 vote	Consensus meeting vote*
Mobility					
1) The child (if age appropriate) is independent with activities of daily livings	2	In			
2) The child's ability to walk unaided	1, 2	No consensus	Yes	In	
3) An improvement in gait (walking pattern) perceived by patient or family	1,2	No consensus	No change	In	
4) A symmetrical rotational profile (feet facing forward as the child walks)	1, 2	No consensus	Yes	No consensus	Consensus out (Out)
5) Foot position in standing/in standing frame	1, 2	No consensus	Yes	No consensus	Out
6) The child's ability to walk barefoot	2	No consensus	Yes	No consensus	Out
7) The child's ability to climb the stairs	2	No consensus	Yes	No consensus	In
8) The child's ability to walk with 'splints'	1	In†			
9) The child's ability to undertake independent transfers	1,2	In			
10) The child's tolerance of splints	1,2	In†			
11) Reduction in child's use of splints	1, 2	No consensus	Remove‡		
12) The child's ability to wear normal footwear	1, 2	No consensus	Yes	No consensus	In†
Musculoskeletal: body form and function					
13) Range of motion in the hip	1	No consensus	Yes	No consensus	In‡§
14) Range of motion in the knee	1	No consensus	Yes	No consensus	In‡§
15) Range of motion in the ankle	1	No consensus	Remove‡		
16) Residual deformity/ stiffness in the hip	1	No consensus	Remove‡		
17) Residual deformity in the foot and ankle	1,2	No consensus	No change	No consensus	In†
18) Coronal and sagittal lower limb alignment (the legs look straight from the front and side)	1	No consensus	Remove‡		
19) Equal leg lengths	1,2	No consensus	No change	No consensus	In
20) Balanced symmetrical spine and pelvic alignment/position	1	No consensus	No change	In	
21) Stable hips that are in joint	1	No consensus	No change	No consensus	Out
22) The foot position when resting on wheelchair plates? (if they have one)	2	No consensus	Remove‡		
23) The child's seating position in their wheelchair?	1,2	In			
Adverse events					
24) Maintaining muscle strength	1	No consensus	No change	In	
25) Absence of pain	1,2	In			
26) Absence of pressure sores	1	In			
27) Good wound healing	1	In			
28) Avoidance of a fracture	1	In			
29) Presence of normal body fat percentage/BMI	2	No consensus	Yes	In	
30) Recurrence of initial need for intervention	1,2	In¶			
31) Recurrent surgery or interventions for same indication/problem	1,2	In¶			
Quality of life: wellbeing, activity, and participation					
32) The child's emotional wellbeing regarding their condition	2	In			
33) The child attends school regularly	2	In			

(*Continued*)

(*Continued*)

Individual clinical outcomes (mapped to ICF domains)	Stage of collection	Delphi stage 1 vote	Change to statement stage 2	Delphi stage 2 vote	Consensus meeting vote*
34) Participation in sport/activities/has a hobby	2	No consensus	No change	In	
35) Socializes with peer group	2	In			

1 = Outcome identified during systematic review.
2 = Outcome identified during questionnaire and interviews process.
*Including changes made to the statements during the consensus meeting.
†Amalgamation of statements 8, 10, 12, 17, 37, and 39.
‡Statement removed after first round of the Delphi due to comments by respondents stating duplication/repetition of the statements.
§Amalgamation of statements 13,14, 40, and 41.
¶Amalgamation of statements 30, 31, 61, and 62.
**Amalgamation of statements 47 and 48.
ICF, International Classification of Functioning, Disability and Health.

are presented in Tables I and II. After discussion and voting during the consensus meeting, 34 of the 41 collected ICO statements appeared in both the ambulatory and non-ambulatory sections. As a result, they were consolidated into 17 ICOs applicable to both groups. The final 28 outcomes, consisting of 24 ICOs (41 minus 17), three Ixs, and one OMT, were grouped within five core outcome domains. The domains are goal setting, general health, clinical examination, mobility and function, and patient-reported outcome measures (PROMs). Investigations are grouped to measure the domains. The GAS is recommended for goal setting.

The final COS, grouped by their domains, is presented in Table V. This forms a goal based 28-point collection tool for the COS of orthopaedic outcomes in ambulatory and non-ambulatory SD.

Discussion

COMET methodology was used to develop a COS to evaluate orthopaedic interventions in SD, define a minimum duration of follow-up, and set timepoints when the COS should be used. At each stage, new outcome domains were identified emphasizing the importance of having an international group inclusive of different stakeholder groups representing both healthcare professionals as well as patients and their families.

This exercise differs from other COS developments that focus on specific body parts, such as the leg and foot in clubfoot. Due to the heterogenicity of SD and the significant spectrum of clinical presentation, this project faced unique challenges. For example, the COS needed to encompass a wide range of presentations and functional goals that evolve as the child grows.

Due to difference in presentation and level of involvement the final product is a goal-based set of outcomes which are centred primarily on functionality of the individual child.

The COS is recommended for use alongside the GAS to ensure that interventions for children with SD are guided by clear, well-defined goals. The COS also assesses whether the goal has been achieved and importantly whether it has been maintained.

In developing the COS, children were classified as ambulatory or non-ambulatory, acknowledging that there is some transition between these groups over time. This approach ensured that relevant outcomes were assigned appropriately, reflecting variations based on each child's functional ability and potential.

During the consensus meeting, the steering group deliberated on statements that had not achieved consensus within their respective domains. Discussions for each statement focused on three key aspects: 1) ensuring the COS remains goal-oriented; 2) emphasizing that orthopaedic interventions in SD aim to support or improve function; and 3) acknowledging that this represents a minimum set of outcomes for reporting while recognizing that other assessment tools, such as gait analysis, add valuable information for specific cases but may not be universally accessible.

Statement 21, 'Stable hips that are in joint' for the ambulatory child did not reach consensus during the Delphi stage. The steering group debated this during the final consensus meeting and agreed that having hips in joint and stable is not always a necessary or realistic goal. Instead, the emphasis should be on symmetry to enable a balanced standing or seating position for optimal function. However, this conflicted with statement 49 for the non-ambulant child, 'Balanced pelvis (both hips dislocated or both hips enlocated)', which had reached 'consensus in' during the Delphi stage. After discussion, all steering group members (12/12) agreed that statement 47, 'Balanced symmetrical spine and pelvic alignment/position', could be used instead, without altering the core meaning of statement 49.

A further area of discussion among the steering group was that of the foot and ankle. There were numerous statements (statements 8, 10, 12, 17, 37, and 39) pertaining to the foot and ankle including splints, footwear, range of motion (ROM), and imaging. From the parents' perspective, it was felt that the child not being able to wear 'off the shelf' footwear was important.

The final decision from the steering group was to merge these statements into one: 'No foot or ankle deformity that interferes with splint use, footwear and/or function including standing frame use'. Similarly with statements surrounding the ROM of the hip and knee, when considering function and the ROM that is needed, the statements were combined and rephrased as 'Passive ROM of hip and knee for function and personal care'.

Table III. The non-ambulatory child: the collected individual clinical outcomes, stage of collection, and voting.

Individual clinical outcomes (mapped to ICF domains)	Stage of collection	Delphi stage 1 vote	Change to statement stage 2	Delphi stage 2 vote	Consensus meeting vote*
Mobility					
36) Child's ability for independent transfers	1,2	In			
37) The child's tolerance of splints	1, 2	No consensus	No change	No consensus	In†
38) Reduction in the child's use of splints	1,2	No consensus	Remove‡		
39) Child's ability to wear normal footwear	1, 2	No consensus	Yes	No consensus	In†
Musculoskeletal: body form and function					
40) Range of motion in the hip	1	No consensus	Yes	No consensus	In§
41) Range of motion in the knee	1	No consensus	Yes	No consensus	In§
42) Range of motion in the ankle	1	No consensus	Remove‡		
43) Residual deformity in the hip	1	No consensus	Remove‡		
44) Residual deformity in the knee?	1,2	No consensus	Remove‡		
45) Residual deformity in the foot and ankle	1	No consensus	No change	No consensus	Out
46) Equal leg lengths	1, 2	No consensus	No change	No consensus	Out
47) Balanced symmetrical spine and pelvic alignment/position	1	No consensus	No change	In¶	
48) Stable hips that are in joint	1	No consensus	No change	No consensus	Out
49) Balanced pelvis (both hips dislocated or both hips enlocated)	1	No consensus	No change	In¶	
50) Child's ability to use a standing frame?	1, 2	No consensus	Yes	In	
51) Child's ability to crawl/ bottom shuffle?	1, 2	No consensus	Yes	In	
52) Foot position in standing/when using standing frame?	1, 2	No consensus	Yes	No consensus	Out
53) Foot position when resting on wheelchair plates?	2	No consensus	Yes	No consensus	In
54/0 Child's seating position in wheelchair?	1,2	In			
Adverse events					
55) Maintaining muscle strength	1	No consensus	No change	No consensus	In
56) Absence of pain	1,2	In			
57) Absence of pressure sores	1	In			
58) Good wound healing	1	In			
59) Avoidance of a fracture	1	In			
60) Presence of normal body fat percentage/BMI	2	No consensus	Yes	In	
61) Recurrence of initial need for intervention	1,2	No consensus	No change	No consensus	In**
62) Recurrent surgery or interventions for the same indication/problem	1,2	No consensus	No change	No consensus	In**
Quality of life: wellbeing, activity, and participation					
63) The child's emotional wellbeing regarding their condition	2	In			
64) The child (if age appropriate) is independent with ADL	2	In			
65) The child attends school regularly	2	In			
66) Participation in sport/activities/has a hobby	2	In			
67) Socializes with peer group	2	In			

1 = Outcome identified during systematic review.
2 = Outcome identified during questionnaire and interviews process.
*Including changes made to the statements during the consensus meeting.
†Combination of statements 8, 10, 12, 17, 37, and 39.
‡Statement removed after first round of the Delphi due to comments by respondents stating duplication/repetition of the statements.
§Combination of statements 13, 14, 40, and 41.
¶Combination of 47 and 48.
**Combination of statements 30, 31, 61, and 62.
ADL, activity of daily living; ICF, International Classification of Functioning, Disability and Health.

Table IV. Investigations and outcome measurement tools: Delphi stage votes.

Investigation	Delphi stage 1 vote	Changes to statement after stage 1	Delphi stage 2 vote	Consensus meeting vote*
1) Achieving the goals of surgery as agreed with the parents/carers[17]	In			
2) Gait analysis[18]	No consensus	No change	No consensus	Out
3) EMG[19]	No consensus	No change	Out	
4) Plain radiographs of the pelvis[3,4,20,21]	No consensus	Yes	In	
5) Plain radiographs of the spine[3,4,20,21]	No consensus	Yes	In	
6) Measurements of the lordosis and/or scoliosis and/or kyphosis[3,4,20,21]	No consensus	Remove†		
7) Plain radiographs of the foot and ankle	No consensus	No change	No consensus	Out
8) Measurement of Reimers migration percentage[3,4,20,21]	No consensus	No change	No consensus	Out
9) Other measurements of hip morphology such as Shenton line, acetabular index[3,4,20,21]	No consensus	No change	No consensus	Out
Outcome measure tool	**Delphi round 1 vote**	**OMT added at round 2**	**Delphi round 2 vote**	**Consensus meeting vote**
1) Hoffer Functional Ambulation Scale[22]	Out			
2) Medical Research Council (MRC) grading[23]	No consensus		No consensus	Out
3) Dimeglio et al[24]	Out			
4) Pirani[25,26]	No consensus		No consensus	Out
5) Classification system of Scott et al[27]	Out			
6) Grading system of Carvalho, Neto, and Dias[28]	Out			
7) Criteria of Dias et al[29,30]	Out			
8) Criteria of Kling et al[31]	Out			
9) Grading system of Legaspi et al[32]	Out			
10) ICFSG and Bensehel[33]	Out			
11) Quality of life PODCI[34]	No consensus		No consensus	Out
12) PedQL[35]	No consensus		No consensus	Out
13)		The Activity Scale for Kids (ASK) Score[36]	No consensus	Out
14)		Goal Attainment Scale in Rehabilitation (GAS)[17]	No consensus	In
15)		Spina Bifida Hip Questionnaire (SPHQ) measuring physical function in children with dislocated hips[37]	No consensus	Out
16)		Spina Bifida Paediatric Questionnaire – quality of life[38]	No consensus	Out
17)		The F words Agreement[39]	No consensus	Out

*Including changes made to the statements during the consensus meeting.
†Statement removed after first round of the Delphi due to comments by respondents stating duplication/repetition of the statements.

Barefoot walking was a topic of debate within the group. Some members felt it should not be encouraged due to altered sensation in SD and the associated risk of injury. However, parents and physiotherapists on the panel emphasized the importance of allowing independence from splints at home. Ultimately, the statement was voted 'consensus out' by a margin of nine to three, with the primary focus placed on maintaining walking ability in general.

The child's ability to manage steps was considered important for accessibility and navigating kerbs when outdoors, and therefore statement 7, 'The child's ability to climb stairs independently', was rephrased as 'The child has maintained their ability to climb a few steps for access'.

The use of standing frame was debated, and it was concluded that its use was important for a number of reasons including bone health, digestion, and participation/inclusion.

Table V. The Spinal Dysraphism Core Outcome Set assessment form.

All children	Ambulatory child	Non-ambulatory child
Goal setting (goal attainment score is recommended)		
Goals of intervention achieved as agreed with the patient/carer		
Goal of intervention maintained		
General health		
Maintenance of muscle strength		
Absence of pain		
Absence of pressure sores		
Good soft-tissue healing		
No history of pathological fracture since last COS collection		
Maintenance of healthy weight for the individual		
Clinical examination		
Optimal pain-free range of motion of hip and knee for function and care	No leg length discrepancy or malalignment that affects function	
Balanced symmetrical spine and pelvic alignment and position		
No foot or ankle deformity that interferes with splint use, footwear and/or function including standing frame use.		
Mobility and function		
The child is able to undertake independent transfers	The child has maintained their walking potential	The child has maintained their ability to use a standing frame.
The child sits comfortably in their wheelchair (if they have a wheelchair)	The child has maintained their ability to climb a few steps for access	The child has maximized their potential to move independently e.g. crawling or bottom shuffling.
		The foot position allows safe and comfortable positioning on wheelchair plates
PROMs		
The child is independent with age-appropriate activities of daily living	An improvement in gait (walking pattern) perceived patient or family	
The child maintains good emotional wellbeing with regards to their condition		
The child attends school regularly		
The child participates in sport or activities/has a hobby		
The child socializes with peer group		
Investigations		
Plain radiographs of the pelvis		
Imaging of the spine to assess deformity (EOS or plain radiographs)		

Timing of core outcome set collection: one year after any intervention. Three standard points of childhood: starting school, end of primary school, and skeletal maturity.
Collected by the lead healthcare professional of the multidisciplinary team.
Using yes/no responses.
COS, core outcome set; PROMs, patient-reported outcome measures.

The inclusion of the adverse events domain under general health in the final COS was deemed essential for outcome reporting following an orthopaedic intervention. This was primarily to assess the potential impact and risks associated with the intervention and to provide valuable insights that can support healthcare professionals and families in making informed decisions for future care.

The key strength of this study lies in its rigorous adherence to the established protocol, its international collaboration, and the comprehensive multidisciplinary contributions, including input from patients and their families. The limitations include the smaller number of participants involved in the Delphi process relative to other core outcome set developments. This is attributable to the rarity and clinical complexity of SD and the inherent difficulties in identifying and recruiting stakeholders with sufficient expertise and experience in this highly specialized area. As with any COS development, they are inherently dependent on the engagement and representativeness of the participating stakeholders and the uptake of the research community.

The COS serves as a minimum set of outcomes that should be reported in all prospective clinical research

to enhance consistency and comparability. Researchers are encouraged to adopt this COS while incorporating additional available outcomes appropriate for a specific patient.

In conclusion, this study developed a COS for orthopaedic interventions in SD, intended for use by all practitioners managing this condition. It comprises 28 outcome parameters, encompassing clinical examination, mobility and functional assessment, PROMs, and investigations. The use of this COS as a minimum set of outcomes to be reported among all children who are undergoing orthopaedic interventions in SD will facilitate meaningful comparisons between patient groups and interventions and the integration of clinical findings into research.

Social media

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Supplementary material

Appendix 1 shows the results from the Delphi round 1, while Appendix 2 shows the results from Delphi round 2. These include respondent background, consensus criteria, analysis of participant response to the outcomes in relation to the ambulatory and non-ambulatory status; analysis of the importance of investigations and outcome measurement tools analyzed by healthcare professionals; healthcare professional responses on optimal follow-up and core outcome set collection.

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Ethical review statement

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