Development of a family-centred core outcome set for infants with developmental dysplasia of the hip treated with a brace

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Aims

This study aims to define a set of family-centred core outcomes for infants undergoing brace treatment to facilitate consistent reporting for future high-quality research.

Methods

Family-centred outcomes will be identified through a literature review and a scoping survey involving key stakeholders, including parents, healthcare professionals, and researchers. These outcomes will then be rated for their perceived importance in a two-stage modified Delphi process with the same stakeholders. Finally, a consensus meeting will be held to establish the final core outcome set (COS).

Conclusion

The impact of brace treatment on the family is profound, but seldom considered in randomized controlled trials. This COS can independently standardize reporting on the family's experience, and potentially become part of a broader COS for developmental dysplasia of the hip in infants undergoing brace treatment.

Take home message

- This protocol is for the development of a family-centred core outcome set (COS) for developmental dysplasia of the hip (DDH) in infants undergoing brace treatment.
- This set can independently standardize reporting on the family's experience, and potentially become part of a broader COS for DDH in infants undergoing brace treatment in the future.

Introduction

Developmental dysplasia of the hip (DDH) encompasses a range of abnormalities affecting children's hip joints, from mild dysplasia to complete dislocation. It occurs in 1% of children. If left untreated or unsuccessfully treated, DDH can lead to premature arthritis and early joint replacement. For infants diagnosed with DDH, nonoperative splinting with a brace is the cornerstone of management.[1] However, there is persistent clinical uncertainty regarding several aspects of brace treatment in these infants. A Cochrane review on splinting for nonoperative management of DDH in children under six months highlighted a lack of randomized controlled trial (RCT) evidence in this area.[2] The review identified key uncertainties requiring further high-quality research, including the effectiveness of splinting for stable dysplastic hips, the optimal timing to begin splinting, the most suitable type of splint to use, and whether a 'weaning' process is necessary when discontinuing splint use.[2]

The wide range of reported outcomes across RCTs complicates the synthesis of results and hinders subsequent clinical decision-making for brace treatment in infants with DDH. Despite the heterogeneity of outcomes reported in trials concerning brace treatment for infants with DDH, very few studies have focused on family-centred outcomes. Families have expressed the

profound impact brace wearing has on the family unit.[3]

A core outcome set (COS) is defined as "the minimum that should be measured and reported in all clinical trials of a specific condition and could also be suitable for use in other types of research and clinical audit."[4,5] This structured approach enhances research efficiency by reducing bias in outcome reporting and ensuring consistency in outcomes across various research studies. This enables the synthesis of findings to bolster available evidence and guide best practices. At present, no COS for DDH exists. The development of a COS for DDH in infants undergoing brace treatment is needed. It is essential that there is a family-centred component to this COS, as the impact of brace treatment on the family unit is significant,[6,7] but rarely considered in the context of RCTs. Understanding the impact of treatment decisions on the family unit is likely to have implications for parent satisfaction, trust, and compliance.[7]

The aim is to create a family-centred COS for infants with DDH undergoing brace treatment, which can be embedded within future high-quality research.

Methods

Setting

This study will mostly take place online; however, a final consensus meeting will be held at a suitable location, which may be in person, online, or hybrid.

Participants

Key stakeholder groups are represented at different stages of the process as appropriate to ensure all views are considered. As per the Core Outcome Measures in Effectiveness Trials (COMET) guidelines, healthcare professionals, researchers, and patient representatives (in this context families) are included in the decision-making process.[5]

- Caregivers (parents or legal guardians) of children with DDH.
- Clinicians involved in DDH care including surgeons, nurses, and physiotherapists.
- DDH researchers.

International participation is planned. All participants will be required to understand English and have access to the relevant technology (digital device capable of loading and completing the online survey, as well as email) to take part.

Identification of key outcomes

Relevant family-centred outcomes will be identified through a literature review and a scoping survey involving parents or legal guardians of children with DDH, as well as clinicians involved in DDH care. Initially, we planned to conduct in-depth qualitative interviews with families to formulate an inclusive list of outcomes. However, upon reviewing previous qualitative studies conducted among families of infants undergoing brace treatment, we determined that additional qualitative work would be unlikely to yield substantial new information. Consequently, potentially relevant outcomes will be identified from existing literature, and key stakeholders will have the opportunity to suggest additional outcomes through a scoping survey.

Literature review: A literature review is planned to provide an overview of relevant qualitative work identifying family-centred outcomes during DDH treatment, aiming to create an inclusive list of such outcomes. A scoping review methodology will be used to address the aim. The Joanna Briggs's Institute (JBI) guidance document for the conduct of scoping reviews[8] and the Preferred Reporting Items for Systematic reviews and Meta-Analyses (PRISMA) extension for scoping reviews (PRISMA-ScR) checklist[9] will be used. The PRISMA-ScR was developed according to guidance from the Enhancing the QUAlity and Transparency Of health Research (EQUATOR) Network for the development of reporting guidelines.[10]

A PEO (Population; Exposure; Outcome) structure is used to define the research question. The search strategy is predefined with a systematic approach to extract relevant studies. The search strategy is outlined in Table I.

Three electronic databases have been identified, and the search strategy will be used for each of them. These are MEDLINE, CINAHL, and ProQuest. Three key articles were identified, including a PhD thesis, and the search strategy sensitivity has been tested to ensure their detection.

We will include any articles exploring family experiences of brace treatment for DDH. This may include, but is not limited to, published qualitative work, theses, and dissertations. The inclusion criteria are any articles exploring the family perceptions or experience of brace treatment for DDH. The exclusion criteria is articles which are specific to surgical treatments of DDH only, and will not be considered. Electronic databases will be limited to English-language and human studies.

Two reviewers (both clinician researchers (JC, OO)) will independently screen identified articles. Any disagreements will be resolved through discussion. Included studies will have the year of publication, study design, study location, study size, and their sample characteristics extracted.

A narrative review will be completed identifying any potential family-centred outcomes for infants undergoing brace treatment for DDH. No critical appraisal will be conducted, and there will be no data synthesis.

Scoping review: We will supplement the review with a survey among families and clinicians to identify any additional outcomes. An online survey will be generated. Participants will be provided with the participant information sheet. Once consent is obtained, participants will be asked some eligibility questions, their role, and basic demographic information. Participants will also be asked to provide a list of family-centred outcomes which they consider relevant for inclusion in future research into a free-text box. The format and wording of all materials presented to all participants will be pre-approved by patient representatives to ensure suitability. The survey will be open for two weeks.

Results will be analyzed by the study steering committee (composed of two clinician researchers and two parent representatives). Any similar outcomes will be grouped. All suggested outcomes will be included.

Delphi consensus exercise

The inclusive list of family-focused outcomes generated from the literature review and the scoping survey will be presented to key stakeholders for rating through a two-round Delphi survey.

Following the guidelines within the COMET initiative handbook,[5] the decision regarding sample size in a Delphi consensus exercise is not based on statistical power and

Table I. Search strategy.

Variable	Search term
Population	"developmental dysplasia of the hip" OR "DDH" OR "CDH" OR "congenital dysplasia of the hip" OR "hip dysplasia"
Exposure	"brace" OR "splint" OR "harness" OR "Pavlik harness" OR "removable rigid splint"
Outcome	"family-centred outcomes" OR "family focused outcomes" OR "family experiences" OR "patient experience" OR "parent experience" or "experiences of parents"

is primarily a pragmatic choice. There are limited resources available to provide guidance. Sample sizes have been chosen to ensure that a broad array of opinions can be gathered, ensuring that the target is ambitious but achievable.

Delphi round 1: An online Delphi consensus exercise with the identified outcomes will be generated, and reviewed by the study steering group to ensure it is easy to understand and accessible to families.

The exercise will take approximately 15 to 20 minutes to complete. Participants will be shown a participant information sheet, and asked to provide consent, confirm eligibility, their role (family, researcher, or clinician), and basic demographic data. Participants will be asked to take part in both stages of the Delphi exercise.

Participants will be presented with each of the outcomes identified and asked to rank them on a nine-point Likert scale. The scoring system is well established, with 1 to 3 signifying an outcome of limited importance, 4 to 6 demonstrating some importance but not essential, and 7 to 9 being of greater importance.[5,11,12] This framework is recommended by the Grading of Recommendations Assessment, Development and Evaluation (GRADE) working group.[13,14] Equal influence is granted to each key stakeholder in the scoring procedure.

Respondents will also be invited to submit additional outcomes and suggest any refinements to the existing outcomes via a free-text box.

The survey will remain open until at least 75 responses have been collected. Reminder emails will be sent after two weeks if 75 responses have not been achieved.

Delphi round 1 analysis: At the close of the first iteration of the Delphi, the study steering group will consider any additional outcomes and suggested refinements. There will be a period of three weeks between each round to allow for response analysis and any amendments to online surveys.

Each outcome will be summarized, with the percentage of participants who rated the outcome as of limited importance,[1,2,4] some importance but not essential,[5–7] and greater importance.[8–10] The breakdown of ratings will be for the group as a whole and for individual stakeholder groups.

Outcomes which meet the "consensus in" or "consensus out" definitions (as specified above) for all stakeholder groups in round one will be presented, but excluded from rating in round two. If one stakeholder group achieved consensus but another did not, then the outcome will be included.

Delphi round 2: The Delphi round 2 will be sent out to respondents to the first round of the consensus process. Each participant will be asked to take part in both rounds of the Delphi exercise unless they withdraw their consent to participate and no longer wish to take part. If this is the case, they will not be sent any further surveys. The survey will again take approximately 15 to 20 minutes to complete.

The results collected in round 1 will be presented to participants as detailed above. Any necessary changes to outcomes will be made based on comments from round 1 and will be clearly signposted to participants.

Participants will be asked to re-rate the outcomes with the knowledge of the group responses. For round two, there will be no free-text box available.

The exercise will remain open for two weeks. An email reminder will be sent if no response is received after one week of non-completion, and a second after ten days. We anticipate an attrition rate of up to 20%, which is deemed acceptable as laid out in the COMET initiative handbook.[5] If 60 responses are not achieved, the survey will remain open for longer if it is felt this is likely to achieve further responses.

Delphi round 2 analysis: Each outcome will be classified as "consensus in", "consensus out", or "no consensus" based on the predefined consensus criteria. The attrition rate will be recorded.

Outcomes that reach consensus (either in or out) during the survey will be forwarded to the final consensus meeting for confirmation. Outcomes which do not research consensus in all stakeholder groups during round one and two will be forwarded to the final consensus meeting for consideration.

Final consensus meeting

The final consensus meeting will contain at least 24 participants, as suggested in the COMET handbook.[15] The consensus meeting will be independently chaired. The meeting will include representatives from all stakeholder groups with a minimum of five parents, five researchers, and five clinicians. The data from the two-round Delphi consensus exercise will be emailed to participants in advance of the meeting.

The consensus meeting will follow a structured agenda beginning with a summary presentation of the results, followed by group discussion. Anonymous scoring will be conducted for outcomes that have not achieved consensus, culminating in the formal endorsement of the final COS.

Consensus definition

The GRADE guidelines[16] recommend evaluating consensus for each outcome by measuring the percentage of participants who rate it within a predefined range. These guidelines will be used to define consensus in this study. The same consensus definition will be applied for both the Delphi process and the final consensus meeting.

"Consensus in" for inclusion in the COS will be indicated by > 70% of the group scoring the outcome between 7 and 9 (greater importance), with < 15% of the group scoring the outcome between 1 and 3 (limited importance).

"Consensus out" for exclusion in the COS will be indicated by > 70% of the group scoring the outcome between 1 and 3 (limited importance), with < 15% of

the group scoring the outcome between 7 and 9 (greater importance).

This definition of consensus has been used by many COS developers, and is well established.

Discussion

At present, no COS for DDH exists. The development of a COS for DHH in infants undergoing brace treatment is needed. It would enable consistent and meaningful reporting, facilitate comparisons in future clinical trials, and aid in establishing gold-standard treatment pathways for these children.[17] It is essential that there is a family-centred component to this COS as the impact of brace treatment on the family unit is substantial,[6,7] but rarely considered in the context of RCTs. The James Lind Alliance found a significant disparity between the priorities of clinicians and researchers and those of patients and caregivers,[18] it is important that parents are considered when gold-standard treatment pathways are determined. Understanding the impact of treatment decisions on the family unit is likely to have implications for parent satisfaction, trust, and compliance.[7] Additionally, when interpreting research, it is important to recognize that if the clinical effects are positive, but the magnitude of the benefit is small, the non-clinical effects on the family may hold greater significance for policy-making and implementation.

This protocol is for the development of a family-centred COS for DDH in infants undergoing brace treatment. This set can independently standardize reporting on the family's experience, and potentially become part of a broader COS for DDH in infants undergoing brace treatment in the future.

Social media

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J. Craven: Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Project administration, Writing – original draft.
O. O'Malley: Data curation, Formal analysis, Writing – review & editing.
D. C. Perry: Conceptualization, Methodology, Project administration, Writing – review & editing.

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Data sharing

The data that support the findings for this study are available to other researchers from the corresponding author upon reasonable request.

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