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PROTOCOL

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Comparison of clinical and radiological outcomes for the anterior and medial approaches to open reduction in the treatment of bilateral developmental dysplasia of the hip: a systematic review protocol

Edward Alan Jenner^{1*} , Govind Singh Chauhan¹, Abdus Burahee^{2,3}, Junaid Choudri², Adrian Gardner^{2,3} and Christopher Edward Bache¹

Abstract

Background Developmental dysplasia of the hip (DDH) affects 1–3% of newborns and 20% of cases are bilateral. The optimal surgical management strategy for patients with bilateral DDH who fail bracing, closed reduction or present too late for these methods to be used is unclear. There are proponents of both medial approach open reduction (MAOR) and anterior approach open reduction (AOR); however, there is little evidence to inform this debate.

Methods We will perform a systematic review designed according to the Preferred Reporting Items for Systematic Review and Meta-Analysis Protocol. We will search the medical and scientific databases including the grey and difficult to locate literature. The Medical Subject Headings “developmental dysplasia of the hip”, “congenital dysplasia of the hip”, “congenital hip dislocation”, “developmental hip dislocation”, and their abbreviations, “DDH” and “CDH” will be used, along with the qualifier “bilateral”. Reviewers will independently screen records for inclusion and then independently extract data on study design, population characteristics, details of operative intervention and outcomes from the selected records. Data will be synthesised and a meta-analysis performed if possible. If not possible we will analyse data according to Systematic Review without Meta-Analysis guidance. All studies will be assessed for risk of bias. For each outcome measure a summary of findings will be presented in a table with the overall quality of the recommendation assessed using the Grading of Recommendations Assessment Development and Evaluation approach.

Discussion The decision to perform MAOR or AOR in patients with bilateral DDH who have failed conservative management is not well informed by the current literature. High-quality, comparative studies are exceptionally challenging to perform for this patient population and likely to be extremely uncommon. A systematic review provides the best opportunity to deliver the highest possible quality of evidence for bilateral DDH surgical management.

Systematic review registration The protocol has been registered in the International Prospective Register of Systematic Reviews (PROSPERO ID CRD42022362325).

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Keywords Developmental dysplasia of the hip, Congenital dysplasia of the hip, Congenital hip dislocation, Developmental hip dislocation, DDH, CDH, Bilateral

Introduction

DDH describes a spectrum of abnormalities in the infant's hip, from subluxation to frank dislocation, due to incomplete acetabular and femoral head development [1]. Developmental dysplasia of the hip (DDH) affects 1–3% of newborns and 20% of cases are bilateral [2–4]. Although many cases of DDH spontaneously resolve as the child grows [5] those in whom the hip(s) remains shallow, subluxed, or dislocated will go on to develop gait abnormalities, hip pain, and early onset osteoarthritis [6]. This often requires early hip arthroplasty [7]. Clinical and radiological outcomes for children with bilateral DDH have been reported to be worse than for children with unilateral DDH by some authors [8–10] whereas others have found no difference [11, 12].

The aim of treatment in bilateral DDH is to achieve concentrically reduced hips, without significant deformity or residual dysplasia. If bilateral DDH is detected as a neonate, abduction bracing is attempted, although failure rates are higher than for unilateral disease [8, 13, 14]. Patients who fail bracing proceed to examination under anesthetic and arthrogram, aiming for closed reduction and hip spica. Typically, this is performed before age 6 months. Bilateral DDH represents a significant risk factor for failure of conservative treatment [8, 9] and patients failing closed reduction proceed to open reduction.

Operative options are medial approach open reduction (MAOR) or anterior approach open reduction (AOR). MAOR is performed between 6 and 18 months of age [15]. This approach requires limited soft tissue dissection through a small, cosmetically acceptable, anteromedial incision with minimal blood loss. The anatomical blocks to reduction (capsular constriction, transverse acetabular ligament, ligamentum teres and iliopsoas tendon) are well visualised and released. Both hips are usually operated on at the same sitting and the patient is immobilised in a hip spica for 6–12 weeks postoperatively. Critics suggest that MAOR increases the risk of femoral head avascular necrosis (AVN), prevents the blocks to femoral head reduction from being fully addressed and does not allow capsulorrhaphy [16–18]. Rates of residual dysplasia may also be higher. It has been reported that MAOR may have worse outcomes compared to AOR [16–18]; however, these studies relate to unilateral cases and limited data, specific to bilateral DDH, has been published. The data relating to unilateral disease is itself heterogeneous and contradictory [15, 19, 20].

AOR is usually performed around 12–24 months of age through a bikini line incision via the ilio-inguinal approach. This results in a larger, less cosmetically acceptable scar, more soft tissue dissection, potentially greater blood loss and risks of damage to the lateral femoral cutaneous nerve [21, 22]. Proponents argue that AOR allows all the potential blocks to femoral head reduction to be addressed and capsulorrhaphy to be performed therefore improving outcomes [23]. Pelvic osteotomy can be performed through the same approach and this is usually required when surgery is undertaken after age 2 years [24–27]. Typically, in AOR, one hip is operated on at each sitting with a 6-week gap between surgeries during which the patient is immobilised in a hip spica cast [11, 12]. Some authors have reported single-sitting bilateral surgery in AOR [25]; however, this remains rare.

The choice of AOR or MAOR depends on a number of factors, including the patient's age, the surgeon's training and experience and the perceived advantages and disadvantages of each technique. Both of these surgical management strategies for bilateral DDH have proponents on each side, however, there is limited evidence to inform decision-making. To the best of our knowledge, this will be the first systematic review comparing outcomes for AOR vs MAOR in bilateral DDH.

Aims

Our aim is to establish whether there is a difference in the clinical and radiological outcomes for children with bilateral DDH who have been treated with MAOR compared to AOR. We will examine a range of clinical and radiological outcome measures and if possible perform a quantitative analysis. We will summarise the evidence available and give recommendations for management. This will help to inform decision-making in the management of bilateral DDH.

Design and methods

This protocol has been designed according to the Preferred Reporting Items for Systematic Review and Meta-Analysis Protocol (PRISMA-P) [28, 29]. The design and method have been formed through discussion between experts in the management of DDH and experts in the methodology of systematic reviews. The protocol has been registered in the International

Prospective Register of Systematic Reviews (PROSPERO ID-CRD42022362325).

Eligibility criteria

Population

Children with idiopathic bilateral developmental dysplasia of the hip undergoing surgical management of both hips.

Exclusion criteria—children with bilateral DDH in whom one hip is managed through harness treatment alone, children with teratologic bilateral developmental dysplasia of the hip, children undergoing revision surgery and surgery for acetabular dysplasia in adolescence.

Intervention

Medial approach open reduction of the hip (MAOR).

Comparison

Anterior approach open reduction of the hip (AOR).

Outcomes

1. Rate and severity of avascular necrosis of the femoral head at the latest follow-up using Kalamchi and MacEwen [30] or Bucholz and Ogden classification [31] or other appropriate scoring system.

and/or

2. Radiological outcome at the latest follow-up using acetabular index measured in degrees, Severin Score [32] or other appropriate scoring system.

and/or

3. Clinical outcomes at the latest follow-up including Modified McKay criteria [33], Children's Hospital of Oakland Hip Evaluation Scale [34], Pediatric Outcomes Data Collection Instrument (PODCI) [35] or other appropriate scoring system.

and/or

4. Prevalence, event rate or time-to-event surgical complications assessed according to the Clavien-Dindo system [36, 37] or other appropriate scoring system.

and/or

5. Prevalence, event rate or time to event of secondary surgery.

Study design

Inclusion criteria—clinical studies, level IV (retrospective case series) and above, with a clear description of the operative management with a set of clinical and/or radiological outcomes included, published in English.

Exclusion criteria—case reports, technical or cadaveric studies, studies without a clear description of the operative management or where this is unobtainable, studies without a clear description of clinical and/or radiological outcomes or where this is unobtainable. Full-text studies not available in English will be excluded.

Search strategy

A search of the electronic medical and scientific databases; PubMed, MEDLINE, the Cochrane Library, Embase, Google Scholar, Web of Science and Scopus will be conducted from the date of first entry until the date of search. The grey and difficult-to-locate literature (including theses and dissertations) will be searched via the Open Grey [38] and Open Access Theses and Dissertations [39] databases. The Medical Subject Headings (MeSH terms) “developmental dysplasia of the hip”, “congenital dysplasia of the hip”, “congenital hip dislocation”, “developmental hip dislocation”, and their abbreviations, “DDH” and “CDH” will be used, along with the qualifier “bilateral”. The search strategy will be developed in Medline and then applied to other databases. An example of the search strategy can be found in Additional file 1. Only full-text studies, published in English will be included. There will be no time limit imposed.

Study selection

Two reviewers (EJ and GC) will independently screen the title and abstract of records for inclusion according to the eligibility criteria. Once preliminary screening has been performed, selected studies will be screened as full text. Researchers will be blinded to each other's decisions. Where there is disagreement a separate reviewer (CEB) will arbitrate. Screening decisions at the full-text stage will be fully recorded. The results of the screening will be presented in a PRISMA flow diagram [29].

Data management

The selected studies will be collated in the Zotero citation management system, screened for duplicates, and exported to Systematic Review Data Repository-Plus [40]. This database will be used to aid data extraction and management. Extracted data will be exported to RevMan software for analysis.

Data extraction

Data will be extracted in a predefined electronic data extraction form. Data on study design, population characteristics, details of operative intervention (intervention and comparison), and outcomes (clinical, radiological, complications and rate of secondary surgery) will be extracted. A summary of intended data

Table 1 Items included for data extraction in selected studies

Data item	Details
Study details	Reference, year of publication, geographical location of study, study design, ethical approval, funding, pre-registered protocol
Population details	Number of patients, age, sex, comorbidities, inclusion/exclusion criteria, duration of follow-up, DDH severity or classification, presence of ossific nucleus
Surgical intervention	Anaesthetic used (incl. Nerve blocks/ spinal), single stage or sequential surgery, Surgical approach, anatomical details of the approach, blocks to reduction addressed and how addressed, presence or absence of ligamentum teres, surgical duration, the volume of blood loss, details of additional procedures (e.g. pelvic osteotomy/femoral osteotomy), method of post-operative immobilisation, duration of post-operative immobilisation
Clinical outcomes including hip-specific outcome scores and general quality of life scores	E.g. Modified McKay criteria [33], Children's Hospital of Oakland Hip Evaluation Scale [34], Pediatric Outcomes Data Collection Instrument (PODCI) [35]
Radiological outcomes including hip and age-specific outcome scores	E.g. rate and severity of AVN (Kalamchi and MacEwen [30] or Bucholz and Ogden classification [31]), acetabular index measured in degrees, Severin Score [32]
Surgical outcomes	Surgical complication rate, surgical complications according to Clavien-Dindo classification, dislocation rate, secondary surgery

items for extraction is shown in Table 1. Four reviewers (EJ, GC, MJC and AB) will be allocated the selected studies and will independently extract data. Each reviewer will be blinded to data extraction. Where possible corresponding authors will be contacted for unreported data. Data will be extracted to a secured anonymised form on Systematic Review Data Repository-Plus and then exported to RevMan for analysis.

Data synthesis

The extracted data will be summarised in a structured table format, grouped and ordered by study design (according to the hierarchy of evidence) or by risk bias if study designs are similar, and including the data items specific to the outcomes of interest. This will help to assess clinical and methodological heterogeneity across the studies and determine the feasibility of performing a meta-analysis. We do not expect the included studies to be of sufficient quality or consistency to allow a meta-analysis to be performed. In this instance, we will follow the Systematic Review without Meta-Analysis (SWiM) guidance [41] and analyse data according to this and the recommendations in the Cochrane Handbook Chapter 12 [42]. Studies will be grouped and tabulated as described. We expect that the key outcome data for radiological and clinical outcomes will be in short ordinal scales (e.g. Severin Score [32]). Where possible we will transform these data to dichotomous outcomes and present this as a relative risk with 95% confidence intervals for MAOR in comparison to AOR. Longer ordinal scales such as the Pediatric Outcomes Data Collection Instrument [35] will be transformed to continuous data. For complications and secondary surgery data we will

transform to an incidence estimate, event rate or time-to-event data. For non-comparative studies, we will transform extracted data as described above and use this to generate a crude estimate of incidence, prevalence or event rate. Where possible we will pool this data using a random effects model as per the recommendation in Murad et al. [43]. Results will be reported according to the guidance in the Cochrane Handbook Chapter 12 [42]. Where sufficient information is available but synthesis cannot be performed a structured reporting of effects will be used. When effect estimates are available without measures of precision an illustrated synthesis of summary statistics will be used. If *P* values are available an illustrated synthesis of *P* values will be used. Where directions of effect are available an illustrated synthesis using vote-counting based on direction of effect will be used.

Meta-bias

We aim to limit publication bias by a thorough and systematic search of the literature including the grey literature as described in the search strategy. Where possible publication bias will be assessed across studies by generation of funnel plots. These will be inspected for asymmetry and analysed via Egger's test [44].

Risk of bias

Randomised trials will be assessed using the Cochrane Risk of Bias 2 (RoB 2) tool [45]. However, included studies are most likely to be non-randomised, observational studies. For comparative studies (cohort or case-control) we will use the ROBINS-I tool to assess risk of bias [46]. For case series, we will use Murad et al.'s method

for evaluating the methodological quality across four domains; selection, ascertainment, causality and reporting [43]. Four reviewers (EJ, GC, MJC & AB) will assess included studies for risk of bias. A separate reviewer (CEB) will resolve disagreements through discussion. A summary figure of the risk of bias analysis will be included in the final manuscript.

Assessment of quality

For each outcome measure a summary of findings will be presented in a table [47] with the overall quality of the recommendation assessed using the Grading of Recommendations Assessment Development and Evaluation approach (GRADE) [48]. This approach uses five factors; risk of bias, inconsistency, indirectness, imprecision and publication bias to assess the quality of evidence and produce a rating of “high”, “moderate”, “low” or “very low”. GRADEpro GDT software [49] will be used to aid decision-making when assessing the quality of evidence.

Discussion and implications of review

Management of bilateral DDH represents a significant challenge for the paediatric orthopaedic surgeon. The aim of treatment is to achieve concentrically reduced hips, without significant deformity or residual dysplasia. The decision to perform MAOR or AOR in patients with bilateral DDH who have failed conservative management is not well informed by the current literature. High-quality, comparative studies are exceptionally challenging to perform for this patient population and likely to be extremely uncommon. A systematic review provides the best opportunity to deliver the highest possible quality of evidence for bilateral DDH surgical management. We are not aware of any systematic reviews that compare the outcomes of MAOR with AOR for bilateral DDH. This study aims to identify whether there are any significant differences in the clinical or radiological outcomes for patients with bilateral DDH surgically treated with MAOR compared to AOR so that surgeons can make better-informed decisions about the management strategy they will offer to patients.

Limitations

We expect that this review will be limited by studies that have a small sample size and have a retrospective, non-comparative study design. We expect result reporting to be heterogeneous and incomplete. These limitations will place all studies at a high risk of bias and therefore limit the quality of evidence that can be derived from the systematic review.

Abbreviations

DDH	Developmental dysplasia of the hip
MAOR	Medial approach open reduction
AOR	Anterior approach open reduction
CDH	Congenital dysplasia of the hip
AVN	Avascular necrosis
PRISMA-P	Preferred Reporting Items for Systematic Review and Meta-Analysis Protocol
PODCI	Pediatric Outcomes Data Collection Instrument
MeSH	Medical Subject Headings
SWiM	Systematic Review without Meta-Analysis
RoB 2	Cochrane Risk of Bias 2
GRADE	Grading of Recommendations Assessment Development and Evaluation approach

Supplementary Information

The online version contains supplementary material available at <https://doi.org/10.1186/s13643-023-02444-6>.

Additional file 1. Search strategy example.

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Authors' contributions

EJ, GC, AB, JC, AG and CEB all made substantial contributions to the conceptualisation, design, background, drafting and editing of this protocol.

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Availability of data and materials

Not applicable.

Declarations

Ethics approval and consent to participate

Not applicable.

Consent for publication

Not applicable.

Competing interests

The authors declare that they have no competing interests.

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