



Review Article

Leg length discrepancy complications from osteotomy procedures in pediatric developmental dysplasia of the hip: A systematic review

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ABSTRACT

Background: In pediatric patients with developmental dysplasia of the hip (DDH), leg discrepancy may occur from treatment complications or from the treatment itself. Surgeons should be mindful that performing osteotomies with the purpose of providing better pelvic joint fit comes with risks of unequal bone growth. This article aimed to systematically review the reported leg length discrepancy (LLD) as a potential complication from osteotomy procedures in surgical treatment of pediatric patients with DDH.

Methods: This systematic review followed the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines. The study protocol was registered on the International Prospective Register of Systematic Reviews. A comprehensive search was performed on PubMed (MEDLINE), Scopus, the Cochrane Library and Europe PubMed Central in March 2022. Studies reporting outcomes of leg length discrepancy after osteotomy was performed were the main inclusion criteria. Quality and risk of bias assessment were performed by individual reviewers.

Results: From existing literatures, a total of eight studies were included in the review. From the data extracted, a total of 94 cases of DDH reported various LLD from 836 published cases with mean incidence of 11.2%. According to the patients' age when the operation was performed, LLD of 2.20 cm was reported from the youngest patient operated on at 1.6 years old and LLD of 1.50 cm from the oldest patient operated on at 18 years old. The median LLD across the included studies was 1.30 cm. Limitations to this systematic review include study risk of bias, LLD reporting inconsistencies and assumptions when extracting the data which might have caused abnormal data distribution. Since no agreement exists regarding how much discrepancy between limb lengths is considered pathological, reports of cases and management of LLD vary widely. These results underline the importance of creating specific criteria to classify LLD severity and recommend appropriate treatment. WC:298.

1. Introduction

Developmental dysplasia of the hip (DDH) is a leading cause of childhood disability. This disorder is responsible for nearly 9% of all primary hip replacements and as many as 29% of those occur in people aged 60 and younger [1]. The variety of developmental hip disorders range from mildly dysplastic, concentrically located, and stable hips to severely dysplastic and dislocated hips. Mild dysplasia may never

manifest clinically or may not manifest clinically until adulthood, whereas severe dysplasia is more likely to present clinically in late infancy or early childhood [1,2].

Due to the advancements in ultrasound imaging, DDH is gaining more attention. Recently, in several European countries, all newborn infants are routinely subjected to ultrasonography to early identify DDH [2,20]. One unintended consequence of routine ultrasonographic screening has been an increase in neonatal treatment, although there is

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still some clinical uncertainty about how to manage these findings [7]. Addressing the benefits and drawbacks of various treatment policies is difficult because of the inconsistency in case definitions, variation in methods of ascertainment, poor quality of most studies, and a lack of evidence from randomized trials.

The earlier DDH is identified, the easier and more effective the treatment would be. The type of treatment is determined by the age of the patient at the time of diagnosis. Non-surgical treatment with a harness or cast is possible when detected early. The primary goal of this treatment is to achieve a stable concentric hip reduction to allow for normal joint development. A harness allows motion while dynamically positioning the hips in flexion and abduction [2,19]. Avascular necrosis (AVN) of the hip is a side effect of this non-surgical treatment [3]. A pelvic osteotomy is required in older children to achieve a stable concentric reduction [4]. When DDH is discovered late, it may be necessary to cut the pelvic bone or the femur to better align the acetabulum and the femoral head so that they develop normally [19]. If DDH is left untreated, it may lead to juvenile coxarthrosis and other serious complications [3,20]. Leg length discrepancy (LLD) may occur from complications of treatment, for example, due to avascular necrosis of the femoral head or from the surgical treatment itself. Surgeons should be mindful that performing osteotomies with the purpose of providing better pelvic joint fit comes with risks of unequal bone growth that will still take place in pediatric population. Research has shown that severe discrepancy between leg lengths may affect gait and performance of daily activities [5].

This study aimed to identify the incidence of LLD that occurs in patients with DDH after osteotomy procedures, and the review results are intended to provide better understanding for surgeons' operative planning and patients' post-operative care.

2. Materials and methods

This systematic review followed the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines (Supplementary material 1 and 2) [16]. The study protocol was registered on the International Prospective Register of Systematic Reviews database before the review was commenced. First, a comprehensive search was performed on PubMed (MEDLINE), Scopus, the Cochrane Library and Europe PubMed Central in March 2022. The search strategy terms are listed in the Supplementary Material.

Inclusion criteria for selected studies were as follows:

- Study design: Randomized clinical trial, controlled clinical trial, case review, case report, systematic review, case series, research studies, research article, follow up study, and cohort studies
- Study group: Patients with developmental dysplasia of the hip aged <18 years old underwent osteotomy surgical procedures
- Interventions: Osteotomy approaches (Salter osteotomy, Dega osteotomy, Pemberton osteotomy, San Diego Osteotomy, Triple innominate osteotomy (Steel, Tonnis, etc.), Ganz (Bernese), Spherical osteotomy, Chiari osteotomy, Shelf procedure (Staheli), and Periacetabular osteotomy
- Comparison: The different osteotomy approaches performed with proximal femoral osteotomy were compared to those performed without proximal femoral osteotomy, which led to leg length discrepancy.
- Outcomes: Incidence in numbers, leg length measurement in centimeters (cm), and incidence during follow-up.
- Language: English

Studies reporting outcomes of leg length discrepancy after osteotomy was performed were included. Exclusion criteria were: non-English language articles, non-pediatric participants, studies that do not include any osteotomy procedure, and commentary reviews or letter to editors. Electronically and printed journals were deemed acceptable.

Three authors (H.M., P.A.S and A.F.I.) independently scanned the studies for potentially eligible titles and abstracts following the inclusion criteria. Duplicates were removed and full texts were obtained from the screened studies after study selection was performed and included studies were consulted with a senior author (Y.D.I.). Any disagreements between authors were resolved through discussion.

Data were extracted with use of the Microsoft Excel (Microsoft, Redmond, WA). Extracted data included basic information regarding the study (author, year, title, type of study, main objective), participant demographics, intervention data (type of operation, age of patient when operation was done), femoral osteotomy performed duration length of follow up, LLD outcome (number of occurrences, leg length discrepancy in cm) and additional data of LLD management done if available. Quality and risk of bias assessments were performed by individual reviewers using the Cochrane RoB tool for randomized studies and Newcastle-Ottawa Quality Assessment Scale for non-randomized studies where appropriate. The extracted data were descriptively and, if available, analytically processed using IBM SPSS Statistics for Macintosh, Version 26 (IBM Corp., Armonk, NY).

3. Results

The PRISMA flowchart [17] for study selection is presented in Fig. 1. The initial search across four medical databases resulted in a total of 126 articles before duplicates were removed. A total of 111 potentially relevant articles were assessed for eligibility criteria and after reading the full texts and applying the eligibility criteria, a total of eight studies were included in the review. Among the included studies, the Newcastle-Ottawa Quality Assessment Scale identified mostly good qualities and only one with fair quality score. Data extracted from the included studies are presented in Appendix (Table 1). We assessed the quality of our systematic review independently using the AMSTAR (Assessing the Methodological Quality of Systematic Reviews) Checklist as critical appraisal tool for systematic reviews with score of high quality (Supplementary material 3) [21].

From the extracted data, a total of 94 cases of LLD reported varied lengths from 836 published cases with mean incidence of 11.2%. The median follow-up time was 5.0 (2.25–15.60) years. The LLD numerical data were tested for normality using Kolmogorov-Smirnov test and its distribution was abnormal. Due to abnormal distribution of the data, the LLD data are presented in median and its minimum and maximum (min-max) length in centimeters (cm) (Table 2).

The shortest length discrepancy was reported 0.63 cm in a child operated on at age 1.7 years and the longest was 5.0 cm in a patient operated on at age 17 years. According to the patients' age when the operation was performed, LLD of 2.2 cm was reported from the youngest patient who was operated on at 1.6 years old and LLD of 1.5 cm from the oldest patient who was operated on at 18 years old. Linear regression analysis of the correlation of LLD with age of operation and type of operation was attempted. However, statistically, due to the abnormal distribution of data, the plotted graph was not presented because of this unmet criterion.

We further grouped the type of operation performed into Pelvic Osteotomy only (PO), Femoral osteotomy only (FO) and combination of both (PO + FO) and presented mean LLD reported from each group of osteotomies (Fig. 2). From the eight studies, both the shortest and longest length discrepancy were reported from patients receiving both PO + FO surgeries.

4. Discussion

Leg length discrepancy (LLD) is one of the possible complications from osteotomy procedures that could lead to walking disturbances in children. Necrotic femoral head or septic arthritis may occur post-operatively, which in pediatric populations are significant risks for osseous growth arrest leading to shortening of the bone [5]. As seen in

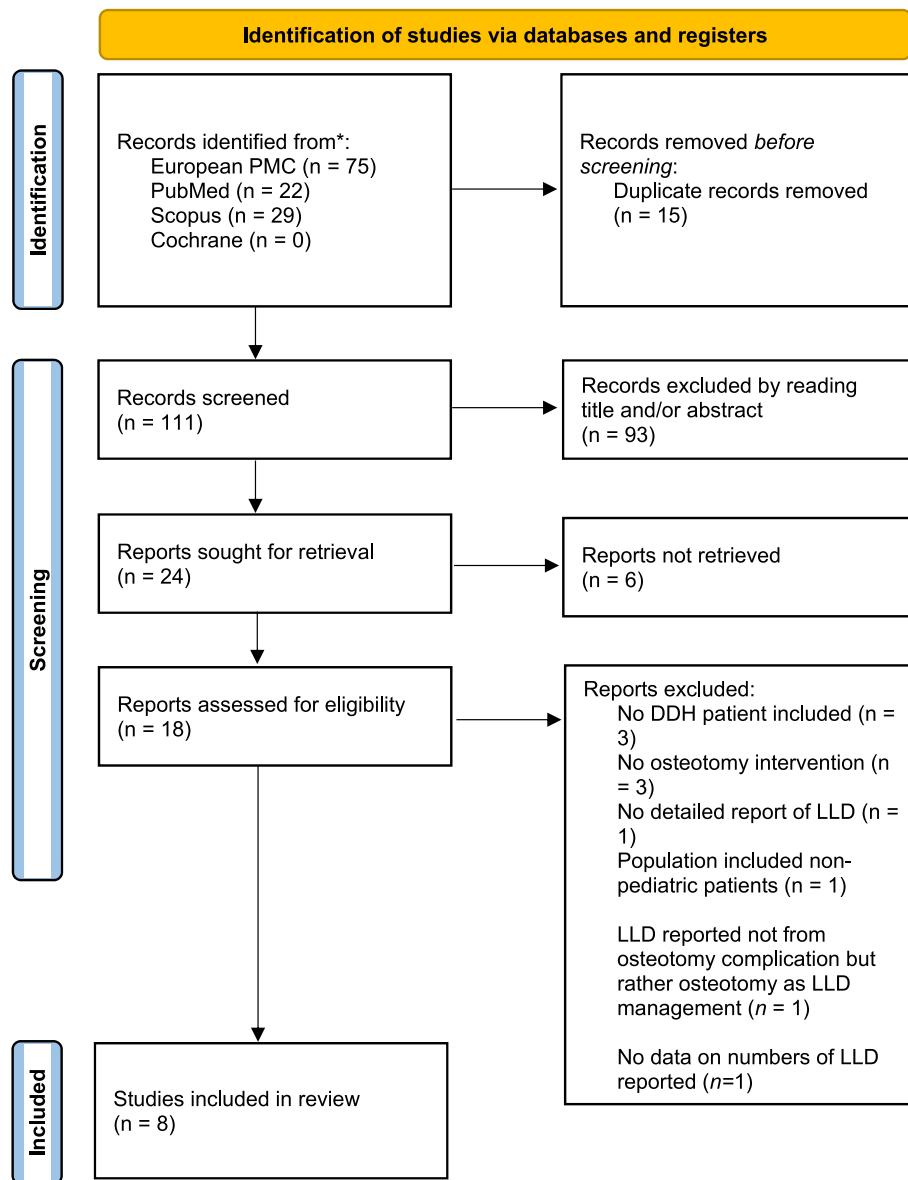


Fig. 1. PRISMA Flowchart for study selection [17].

the number of literatures included in this review, length discrepancy following osteotomy procedure is scarcely reported although its sequelae may impact individual's gait. The incidence of length discrepancy following surgical treatment of DDH has not been reported previously. Alassaf et al. [6] reported the occurrence of LLD in the number of hips affected rather than actual patients. Our reported incidence of LLD in this case assumed these affected hips were from different patients and counted them as separate individuals. In obtaining the true incidence of DDH, Bialik et al. [7] faced some difficulty due to the various methods of diagnosing DDH. Similarly, reporting LLD has not been uniformly done by authors and its methods of measurement are varied. Furthermore, the other seven studies in this review did not specifically describe how they measured the length discrepancy and we assumed they measured it manually, while Yoon et al. [8] recorded the difference by measuring the femoral head height from pelvic radiograph taken antero-posteriorly.

The lack of a criteria or any clear consensus in exact LLD measurement may have caused underreporting of LLD complications from osteotomy surgery. Nakamura et al. [9] reported that no treatments were performed in patients observed with LLD less than 2 cm since the

authors predicted compensation would take place from trochanteric overgrowth as the child grows. Meanwhile, operative outcomes reported by Sokolovsky and Sokolovsky [10] included patients with length differences ranging from 0.50 to 1.50 cm from previous surgical treatment for DDH to undergo corrective rotational osteotomy. Ok et al. [11] reported two patients with LLD of 5.00 cm where one had femoral lengthening procedure and the other had shoe raise to counterbalance the length difference. Epiphysiodesis was the management opted by Inan et al. [5] and Yoon et al. [8] for patients with length discrepancy ranging up to 1.50 cm. From our review, the median leg length difference across all of the included studies was 1.30 cm.

Up to 40% of patients undergoing closed or open reduction still require further surgery from residual dysplasia of the hip [3,19]. In children older than 1.5 years, dysplasia or persistent dislocation due to conservative treatment or reduction failure would require pelvic or femoral osteotomies as the main treatment of choice [12,18]. Femoral osteotomy involves shortening the femoral head which reduces the complication risk of osteonecrosis that may follow from prolonged excessive contact pressure. Meanwhile, pelvic osteotomies, grouped into three major types, alter the acetabulum and are proven to be more

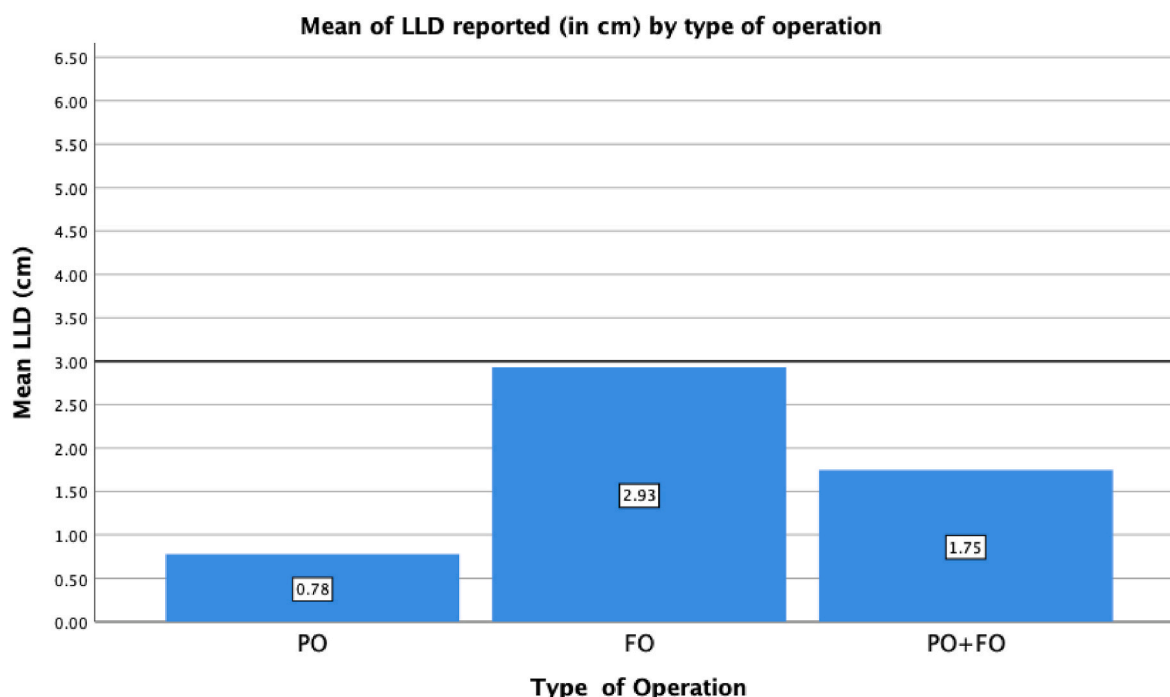


Fig. 2. Mean leg length discrepancy in centimeters reported from each group of osteotomies; FO: Femoral osteotomy; PO: Pelvic Osteotomy.

effective in improving radiological acetabular index compared to femoral osteotomy [13]. As presented in the review results, the mean LLD reported was greatest when patients have received femoral osteotomy alone compared to pelvic osteotomy or the combination of both, with average discrepancy of almost 3.00 cm (Fig. 2). A multivariate analysis was done by Yoon et al. [8] which took into account study participants' types of osteotomies, considering whether they were femoral osteotomy only, pelvic osteotomy only or combination of both and obtained femoral osteotomy as the sole significant risk factor of lower limb overgrowth especially when performed at 2-4 years of age. Spence et al. [13] revealed more than 60% of patients reported complications with severe osteonecrosis (Kalamchi grade III or IV) following femoral derotation osteotomy compared to one in four case reports of patients after pelvic innominate osteotomy. Femoral osteotomy greater than 3.00 cm and complications of avascular necrosis of the femoral head have increased risks of developing LLD [5]. These findings may explain the greater LLD reported in patients undergoing femoral osteotomy only compared to other surgical groups.

5. Conclusions

Osteotomy is considered a final option in DDH treatment stages, reserved for cast and reduction-resistant cases or due to delay in seeking medical treatment. The reporting of LLD as a potential complication following osteotomy procedures in cases of pediatric DDH is still rare. Since no agreement exists regarding how much discrepancy between limb lengths considered as pathological, reports of cases and management of LLD are varied. Factors that may contribute to the occurrence of LLD include femoral osteotomy and patients' age when the operation was performed since these are related to growth arrest.

Limitations in this review include the risk of bias from included studies and differences in reporting LLD between studies. Since reports of LLD from most of the included studies were under-detailed (e.g. reported limb discrepancies in ranges or means of lengths, inconsistent reporting hip number rather than participant number, age of operation performed reported in ranges or means) assumptions were made in order to extract comparable data between studies. In order to conclude LLD incidence in percentages, the extracted data were mainly reported

in means. Due to these assumptions, distribution of data was abnormal, thereby limiting our interpretations.

Regardless of these limitations, this review emphasizes the importance of leg length discrepancies that may follow osteotomy procedures as a potential complication. Furthermore, consensus statements or specific criteria should be developed for more uniform case reporting and management.

Ethical approval

Ethical clearance was not required by the Ethic Committee of Faculty of Medicine, Public Health and Nursing, Gadjah Mada University as this review did not directly involve human nor animal subjects.

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This research received no external funding.

Author contribution

H.M., P.A.S., A.F.I.: research concept, literature search, data analysis, manuscript preparation, drafting the manuscript, reviewing and editing the manuscript, visualizing the data into table; Y.D.I.: research concept, data analysis, manuscript preparation, reviewing and editing the manuscript, visualizing the data into table, supervision of review process.

Research registration number

The study protocol was registered on the International Prospective Register of Systematic Reviews (PROSPERO; CRD42022300411) database prior to start of review.

Guarantor

All of the authors take full responsibility of this study.

Consent

The authors gave consent for publications.
N/A.

Standards of reporting

This systematic review followed the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines. The study protocol was registered on the International Prospective Register of Systematic Reviews database prior to start of review.

Availability of data and material

The authors declare that all data generated or analyzed during this study are included in this published article.

Provenance and peer review

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Registration

Prior to this commencement, this review has been registered the International Prospective Register of Systematic Reviews (PROSPERO; CRD42022300411).

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.ijso.2023.100665>.

APPENDIX**Table 1**

Extracted data summary

Author	Title	Number of participants	Age of operation (years)	Intervention	Length of follow up (years)	Number of LLD reported	Outcome of LLD
Brdar et al. (2013) [14]	Walking quality after surgical treatment of DDH in children	39 patients	age divided into 3 groups Group I: ≤2 Group II: 2-4 Group III: >4	Salter innominate osteotomy + femoral derotation and shortening	Group I: 11.25 Group II: 12.33 Group III: 14.88 (mean)	12/39 patient Group I: 4 patient Group II: 5 patient Group III: 3 patient 9 patient	Group I: 0.63 cm Group II: 1.30 cm Group III: 1.50 cm
Inan et al. (2008) [5]	The correction of LLD after treatment in DDH by using a percutaneous epiphysiodesis	12 patients	2,5 - FVDO @ 1.7 - FS + PIO @ 3.4 - OR + SIO @ 1.8 - OR + FVDO + FS + PIO @ 4 - OR + SIO @ 1.6 - OR + PIO @ 4 - FVDO @ 4.8 - FDO @ 2,5 - SIO @ 10 - FDO + FS @ 3.5	Femoral varus derotation osteotomy + Femoral shortening (OR) @ 1yo - Femoral varus derotation osteotomy - Femoral shortening + Pemberton iliac osteotomy (OR) @ 1yo - Open reduction + salter innominate osteotomy - Open reduction + femoral varus derotation osteotomy + femoral shortening + pemberton iliac osteotomy (OR) @ 1yo - Open reduction + Salter innominate osteotomy - Open reduction + pemberton iliac osteotomy - Femoral varus derotational osteotomy (OR) @ 1.0 yo Femoral derotation osteotomy Salter innominate osteotomy (OR) @ 1.2 yo	Mean: 4.6		1.8 cm 3.3 cm 2.2 cm 2.5 cm 3.3 cm 3 cm 2.5 cm

(continued on next page)

Table 1 (continued)

Author	Title	Number of participants	Age of operation (years)	Intervention	Length of follow up (years)	Number of LLD reported	Outcome of LLD
				- Femoral derotation osteotomy + femoral shortening (OR) @ 1.6 yo - OR + FVDO + FS @ 2 - TPO @ 8 - Open reduction + Femoral varus derotation osteotomy + femoral shortening - Triple pelvic osteotomy - FDVO with shortening ± Salter innominate osteotomy			3.2 cm
Nakamura et al. (2004) [9]	Long-term result of combination of open reduction and femoral derotation varus osteotomy with shortening for developmental dislocation of the hip	9 patients	range: 0.92–4.08 (Mean: 2.08)		Mean: 15.58	3 patient	≤2 cm
Ok et al. (2007) [11]	Operative treatment of developmental hip dysplasia in children aged over 8 years	9 patients	range: 8–17	- Open reduction - Femoral shortening and varus derotation osteotomy ± femoral shortening ± Chiari pelvic osteotomy	Mean: 7.1	2 patient	5 cm
Sokolovsky and Sokolovsky (2001) [10]	Posterior rotational intertrochanteric osteotomy of the femur in children and adolescents use in residual deformity of the femoral head after treatment for developmental dysplasia of the hip.	36 patients (37 hips)	range: 6–18	posterior rotational intertrochanteric osteotomy of femur	Mean: 4.42	6 patient	0.5–1.5 cm
Subasi et al. (2008) [15]	Outcome in unilateral or bilateral DDH treated with one-stage combined procedure	40 patients (51 hips)	range: 3–10	open reduction + salter osteotomy + femoral derotation + shortening osteotomy	Mean: 6.7	4 patient	>1.5 cm
Wang et al (2016) [16]	The comparative, long term effect of the Salter osteotomy and Pemberton Acetabuloplasty on pelvic height, scoliosis and functional outcome	42 patients	range: 1–3	open reduction + capsulorrhaphy + salter osteotomy/pemberton acetabuloplasty	Range: 5–10	42 patient	MEAN: Salter: 0.63 cm Pemberton: 0.23 cm
Yoon et al. (2020) [8]	Overgrowth of lower limb after treatment of developmental dysplasia of the hip: incidence and risk factors in 101 children with a mean follow up of 15 years	101 patients	range: 2–4	femoral osteotomy	Mean: 15	67 patient	1.0–1.5 cm
Alassaf et al. (2018) [6]	Predictors of femoral shortening for pediatric developmental hip dysplasia surgery: an observational study in 435 patients	548 hips Hips with femoral shortening: 119 hips with no femoral shortening: 429	range: 1–8	Open reduction ± femoral shortening ± pelvic osteotomy	Mean: 2.25	3/548 hips	>2 cm

Table 2
Leg length discrepancy in centimeters

LLD in centimeters	(n = 94)
Median (min-max)	1.30 (0.63–5.00)

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