

One stage procedure for developmental dysplasia of the hip in patients aged eight years or older. Systematic review and meta-analysis

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The objective of the present study is to perform a systematic review with meta-analysis of the literature on treatment of developmental dysplasia of the hip (DDH) in patients over eight years of age to provide better understanding of therapeutic strategies and results. Authors carried out a systematic review and metanalysis of the literature on DDH treated in patients aged eight years of age or older. A literature search was carefully performed from June 2019 to June 2020. The articles had to report one stage reconstructive surgical treatment of DDH for patients eight years or older, presenting clinical and radiographic evaluation according to the Tonnis and Severin, and McKay systems. Meta-analysis was carried out using the software metanalyst to perform the pooled effect size Nine studies met the inclusion criteria. They assessed a total of 234 patients and 266 hips. Female patients accounted for 75.7% (eight unknown) and the follow-up ranged from 1 to 17.4 years. The majority of the procedures included an acetabular surgery (93.9%) while femoral shortening was performed in 78%. Acceptable outcomes were found in between 67% (Mckay system) and 91% (Severin system) of the cases. Femoral varus and derrotation shortening associated with redirectional osteotomy of the acetabulum (for those with closed triradiate cartilage) or acetabular redirection/reshaping were the most prevalent combined procedures; this strategy can lead to 60% of acceptable clinical results and 90% of radiographically acceptable results. Therefore, our findings give credence to the recommendation for the treatment of DDH in patients over eight years old.

Keywords : Hip, surgery, dysplasia, treatment.

INTRODUCTION

Neglected or late presentation of developmental dysplasia of the hip (DDH) in patients eight year of age or older is still a worldwide problem, but especially in developing countries^{1,2} or among low socioeconomic groups. Treatment goals aim to achieve a mobile, well-developed, stable and painless hip. These are challenging objectives to achieve due to anatomic alterations during patient growth. The major issues to be addressed are shallow acetabulum with low potential for remodeling, soft tissue adaptation, muscle contractures, high and deformed femoral head¹⁻⁴.

Open reduction associated with pelvic and femoral osteotomies (the so-called one stage procedure) are proposed surgical treatments for late-presenting DDH^{2,3,5,6}. The procedure is complex, technically demanding and only few centers and surgeons have enough experience to perform safely this type of surgery⁴⁻⁶. Although one stage procedure is feasible for treating late-presenting DDH, there is still doubts

about its effectiveness, in addition, conflicting results and only few studies consisting of small sample size were designed to address the problem²⁻⁶.

Single stage surgical treatment of DDH in older children or adolescent can result in major complications such as avascular necrosis of the femoral head, redislocation or subluxation, pain and stiffness^{3,5-7}. Therefore, some authors do not advocate treatment of DDH in patients from eight years of age or older^{2,8}. On the other hand, there are successful reports in which those patients had good outcomes^{2,9-11} despite the difficulties of the surgical procedure. This study is a systematic review with meta-analysis of the literature on treatment of DDH patients over eight years of age to provide better understanding of therapeutic strategies and results.

MATERIAL AND METHODS

Authors carried out a systematic review and metanalysis of the literature on developmental

dysplasia of the hip surgically treated in patients aged eight year of age or older. The systematic review followed the PRISMA guideline and was performed from June 2019 to June 2020.

A literature search was performed using the following electronic resources: MEDLINE/PubMed, Embase, The Cochrane Library, CINAHL, Web of Science, Scopus, and Biblioteca Virtual em Saúde (BVS). The search was carried out using selected terms from the Medical Subject Headings (MeSH), as well as a combination of chosen key words. The strategy was not limited to that electronic search, but also included, google scholar, proceedings and meetings, and citations in the reference list of all selected papers. The key words used were: "hip dislocation, congenital" OR "hip" [All Fields] AND "dislocation" [All Fields] AND "congenital" [All Fields] OR "congenital hip dislocation" [All Fields] OR "developmental" [All Fields] AND "dysplasia" [All Fields] AND "hip" [All Fields] OR "developmental dysplasia of the hip" [All Fields] AND "eight" [All Fields] AND "years" [All Fields] AND "older" [All Fields] AND "therapy" [Subheading] OR "therapy" [All Fields] OR "treatment" [All Fields] OR "therapeutics" OR "therapeutics" [All Fields] OR "surgery" [All Fields]. All the procedures were performed in accordance with the PRISMA guidelines for systematic reviews.

The articles had to report orthopedic one stage reconstructive surgical treatment of DDH for patients eight years or older. The articles had to describe clinical and radiographic evaluation according to the classification systems proposed by Tonnis and Severin (radiographic), and McKay (clinical)¹²⁻¹⁴. These assessments should have been performed preferably in both pre and post-operative periods, or at least in the later one. Exclusion criteria consisted of studies involving neuromuscular disease, connective tissue disorders or hip infection.

The search strategy was performed by two of the authors separately. They took into consideration the title and abstract of the paper to carry out the first evaluation and selection of the studies. After selection based on the exclusion and inclusion criteria, selected studies were obtained in complete text. Hereafter, the articles were read separately by four of the authors to verify whether the paper had met all the established criteria of the systematic review.

Three of the authors collected the data from the studies by means of a standard form. A different author independently revised the extracted data from both the studies and the forms. Analyzed features included (but not limited to) authors, title, publication data, geographic origin, study design, sample size, clinical and radiographic evaluation, patients age and gender, pre-operative condition, detailed intervention (surgical procedure), follow-up, and outcomes.

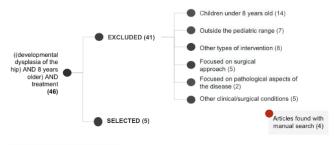
Descriptive statistics was used to present data from all the selected studies, using mean and standard deviation for continuous variable or number and percentages for categorical ones. With regard to the metanalysis, the software MetaXL was used to perform the pooled effect size. Authors analyzed clinical outcomes according to the McKay classification and the analyzed radiographic evaluations using the grade system proposed by Severin. These two outcome measures were put into the metanalysis model to classify results as acceptable or unacceptable. In the McKay classification, results graded as I and II were considered acceptable, while grades III and IV were considered inacceptable. In the Severin classification, result grades I through III were considered acceptable, while grades IV through VI were considered unacceptable. The random effect assessment was used to perform the meta-analysis in order to reduce study heterogeneity. All the statistical tests adopted a significance level of 0.05.

RESULTS

The present study includes articles dealing with surgical treatment of developmental dysplasia of the hip (DDH) in patients aged eight or older. Articles were included irrespective of surgical technique. The summary of the strategy using the proposed terms followed algorithm 1.

Nine studies met the inclusion criteria^{2-5,6,9-11,15}. They assessed a total of 234 patients and 266 hips. Female patients accounted for 75.7% (eight unknown) and the follow-up ranged from 1 to 17.4 years. Only Three studies were prospective (See Table I).

Neglected cases represented 52.6% of the cases, whereas 16.5% consisted of previously operated



FILTERS - Publication date (2000-2020) Language (English)

Algorithm 1 — Selection of the studies according to the proposed terms.

Study	Design	Follow-up (Yrs)	Country	Subjects (Female)	Hips	Age (average)		
Qadir et al. 2018	Retrospective	2	Paquistan	65 (46)	77	8-15 (11.1)		
Ning et al. 2014	Retrospective	3.2-8.9	China	78 (65)	82	8-13 (9.6)		
Yagmurlu et al. 2013	Prospective	2-7	Turkey	9 (4)	9	8-14 (10.2)		
Xu et al. 2010	Retrospective	5-11.5	China	22 (14)	27	7-14 (8.5)*		
El-Tayeby et al. 2009	Retrospective	3.9-5.3	Egypt	16 (8)	19	8-18 (10.6)		
Papavasiliou et al. 2005	Prospective	5-13	Greece	11 (10)	16	10-17 (12.2)		
Inan et al. 2005	Prospective	3.2-4.1	Turkey	8*	8	8-11 (9.8)		
Dogan et al 2005	Retrospective	1-4.2	Turkey	10 (10)	13	9-14 (12.7)		
Wada et al. 2003	Retrospective	5-17.4	Japan	15 (14)	15	8-11 (9.5)		
Total	-	-	-	234 (171)	266	-		
*Included because the minimum age was greater than seven.								

Table I. — Main characteristics of the selected studies.

patients, and in 82 out of 266 (16.5%) cases there were no information about previous procedures.

Operative techniques consisted of one stage surgical procedure, including open reduction of the femoral head associated with acetabuloplasty and femoral shortening, several kinds of reshaping acetabuloplasty (Dega, Pemberton) and partial redirectional osteotomy (Salter operations)^{9,16-21}. Femoral varus or derrotation were performed according to surgeons' judgement based on image studies or clinical features. The goals of femoral surgery were to restore the shenton's line (shortening) and to adjust anteversion to no more than 20 degrees¹⁹.

The majority of the procedures included an acetabular surgery 93.9% (250); among them, dislocation of the entire acetabulum by means of triple (65 out of 126 were Steel type) or doble osteotomies (144; 54.2%144), redirecting/reshaping osteotomies (42 Salter, 23 Pemberton, 1 Dega; 24,8%), and acetabular augmentation (27 out of 40; 15.1%)^{9,16-21}.

Femoral shortening was performed in 78.2% (208) of the cases, and in 10.2% (27) cases it was not possible to define the type of femoral surgery. Cases in which shortening was performed, varisation was associated

in 68 children (32.7%), derrotation combined with varisation in 41 (19.7%), and derrotational osteotomy alone was performed in 79 (29.7%). In cases in which no shortening was carried out, just one was varisation (3.2%), and 21 consisted of varus derrotational osteotomy (67.7%). The need for varus procedure occurred in 54.8% of the cases (131), the need for derrotation in 58.9% (141), and shortening alone in 8.2% (20).

Only four studies classified the hips according to the Tonnis grade system¹². From those reports 55.9% were of low or high luxation (Table II). Postoperatively, all the selected studies classified the results following at least one of both systems: Severin (nine studies) or McKay (seven studies)^{13,14}.According to McKay's classification, acceptable results (I or II) were reported in 59.1% of the hips; according to Severin's system, acceptable results (I through III) were achieved in 80.4% of the hips (Table III).

The meta-analysis was performed for both, clinical and radiographic results using the random effect size to correct for study heterogeneity. The pooled random effect of the clinical results showed that 67% (p<0.001) of the cases can be considered acceptable

Study	Tonnis (preoperative)						
	Ι	II	III	IV			
Qadir et al. 2018	10	14	22	31			
Ning et al. 2014	16	46	10	10			
Yagmurlu et al. 2013	0	0	6	21			
Xu et al. 2010	-	-	-	-			
El-Tayeby et al. 2009	-	-	-	-			
Papavasiliou et al. 2005	-	-	-	-			
Inan et al. 2005	0	0	2	6			
Dogan et al 2005	-	-	-	-			
Wada et al. 2003	-	-	-	-			
Total (%)	26 (13.4)	60 (30.9)	40 (20.6)	68 (35.1)			

 Table II. — Initial radiographic evaluation of the patients according to Tonnis classification system.

Study	Mckay (postoperative)				Severin (postoperative)					
	Ι	II	III	IV	Ι	II	III	IV	V	VI
Qadir et al. 2018	22	44	9	2	38	19	14	6	0	0
Ning et al. 2014	6	8	43	25	21	8	13	6	21	13
Yagmurlu et al. 2013	1	2	4	2	3	2	3	1	0	0
Xu et al. 2010	16	8	3	0	15	12	0	0	0	0
El-Tayeby et al. 2009	10	5	3	1	14	3	2	0	0	0
Papavasiliou et al. 2005	-	-	-	-	4	9	3	0	0	0
Inan et al. 2005	2	3	2	1	0	6	2	0	0	0
Dogan et al 2005	-	-	-	-	9	1	0	2	0	1
Wada et al. 2003	12	1	2	0	7	5	1	2	0	0
Total (%)	69 (29.1)	71 (29.9)	66 (27.8)	31 (13.1)	111 (41.7)	65 (24.4)	38 (14.3)	17 (6.4)	21 (7.9)	14 (5.3)

Table III. — Clinical and radiological results according to McKay and Severin's classification system.

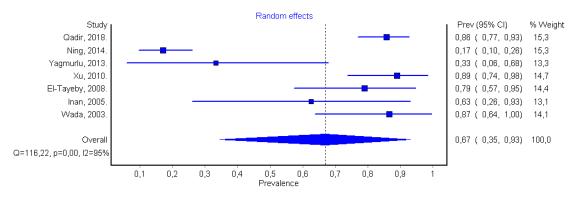
using McKay's criterion. On the other hand, pooled random effect for radiographic results showed 91% (p<0.001) of acceptable results (Graph. 1 and 2).

DISCUSSION

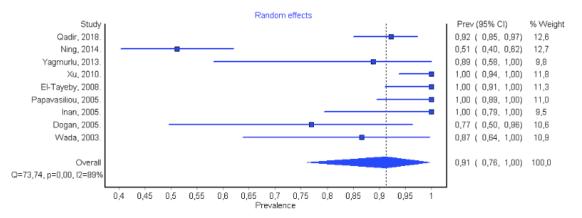
The present findings demonstrate that pediatric orthopedic surgeons can expect 59% of acceptable clinical results (McKay's system) and 80% of radiographically acceptable results (Severin System) when one stage surgery for DDH is performed in patients older than eight years of age. Our meta-

analysis demonstrates that 67% of clinical results can be considered acceptable, while 91% are radiographically acceptable according to the pooled effect (both statistically significant). As clinical results are more important, the surgeon can expect that at least two out of three patients will be satisfied with the treatment outcome.

Our results suggest that the most recommended procedures are open reduction followed by femoral shortening associated with varus and/or derrotation and acetabular osteotomy. Acetabular surgery was needed in almost all patients (94%), being the



Graph. 1 — Random polled effect for clinical results.



Graph. 2 — Random polled effect for radiographic results.

recommended procedures the ones that redirect the entire acetabulum (double or triple osteotomies) in 54% of the cases, followed by redirecting (Salter), reshaping (Pemberton), or augmentation surgeries (Staheli). Femoral shortening was performed in almost all cases (78%), femoral derrotational osteotomy was used in more than half (53%), while varus osteotomy was used in 50% of the patients; at least one of the procedures (varus or derrotation) was performed in 92% of casest.

Especially in subluxated or dislocated cases, femoral shortening is essential for maintaining the reduction of the femoral head without concern of redislocation and/ or excessive pressure on the head, possibly leading to avascular necrosis^{3,11}. It is also clear that as the patient gets older, anteversion and valgus becomes a fundamental alteration to be corrected. Femoral varus or derrotation were performed according to surgeons' judgement based on image studies or clinical features¹⁹. The goals of the femoral surgery were to restore the Shenton's line (shortening) and to adjust anteversion. The amount of shortening reported varied from 1 to 3 cm; the amount of varisation is limited so that a neck shaft angle of at least 90° to 100° is maintained; derrotation was stated as 10-20 degrees^{1,2,6}.

As patients get older, acetabular procedures can vary significantly. Salter redirectional osteotomy is advisable when there is still potential for acetabular remodeling from 18 months to a maximum age of four years^{22,23}. Another prerequisite for single innominate osteotomy is a mild dysplasia because this procedure is unable to adequately correct deformities greater than 15 degrees²². After the age of 8-10 years, the symphysis pubis closure or stiffness limits the mobility of the acetabulum, therefore single innominate osteotomy is not adequate for this purpose^{24,25}. Pemberton, Dega and other reshaping osteotomies were able to correct larger deformities, but they depended on the mobility and opening of the triradiate cartilage which is possible until the ages of 10-12 years^{16,17,24,26}. Double or triple osteotomies are performed independent of either a mobile triradiate cartilage or a mobile symphysis²⁵. Finally, augmentation procedures are ideal for the oldest patients in which the acetabulum is largely dysplastic and cannot cover completely the femoral head^{9,20,21}

Two of the strongest studies had conflicting results and deserve special attention2,3. Ning et al. had good clinical results for about 17% of cases contrasting with Qadir 2018 that presented 86% of satisfactory clinical outcomes^{2,3}. Ning et al. used triple pelvic osteotomy in 75.6% (Salter or Pemberton in the remaining) of their patients while Qadir et al. used triple or double osteotomy in 84.4% (Salter in the remaining). Both studies used femoral shortening associated with derrotation when needed. However Only Qadir et al. used femoral varisation in all their cases. The radiographic assessments showed less impressive differences between these studies, namely 51%³ versus 92%². Those results can indicate that femoral derrotational varus osteotomy associated with triple osteotomy could be the best choice, besides it could also evidence that radiographic results tend to overestimate good results.

The limitation of the present study is attached to the design and pitfalls of the primary papers. Initial classification by the Tonnis grade system as well as other clinical details that could have influenced the results were not provided in most of the studies. Heterogeneity of the patients such as previous procedures, clinical follow-up, results according to age groups, few large samples, and multiplicity of surgical interventions are among several other factors that should be considered when interpreting the results of this metanalysis. The impact of re-operation, for instance, was not evaluated due to lack of consistent data. It should be noted that in the current sample of papers, only one clearly presented a strong negative result³ therefore, the present study should be interpreted with caution due to the tendency of not publishing negative results, a common scientific bias.

One-stage procedure including three types of operations could be applied to reconstruction of the hip in patients older than eight years presenting neglected developmental dysplasia of the hip. Open reduction, femoral shortening associated with varus and derrotation, and dislocation of the entire acetabulum (for those with closed triradiate cartilage) or acetabular redirection/reshaping osteotomies were the most prevalent type of combined procedures. The results of our meta-analysis provide evidence that patients older than eight years of age treated for DDH by varus femoral shortening associated with acetabuloplasty present significant and consistent good outcomes. Using this strategy, pediatric orthopedic surgeons can expect around 60% of acceptable clinical results and around 90% of radiographically acceptable results. Therefore, our findings give credence to the recommendation for the treatment of DDH in patients over eight years old.

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