



Review Article

Hip salvage surgery in cerebral palsy cases: a systematic review[☆]



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ARTICLE INFO

Article history:

Received 6 September 2012

Accepted 14 May 2014

Available online 16 June 2015

Keywords:

Cerebral palsy

Salvage

Hip

Femur

ABSTRACT

Imbalance and muscle spasticity, in association with coxa valga and persistent femoral anteversion, compromises hip development in cases of cerebral palsy and may result in chronic pain and even dislocation. Some of these hips undergo salvage surgery because of the severe impact of their abnormalities in these patients' quality of life. We conducted a systematic review of the literature to compare the results from the main hip salvage techniques applied to these individuals. The literature search focused on studies that evaluated results from hip salvage surgery in cases of cerebral palsy, published from 1970 to 2011, which are present in the Embase, Medline, PubMed, Cochrane Library and SciELO databases. Although the results were not statistically comparable, this systematic review demonstrates that hip salvage surgery should be indicated after individual evaluation on each patient, due to the wide spectrum of presentations of cerebral palsy. Therefore, it seems that no surgical technique is superior to any other. Rather, there are different indications.

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Cirurgias de salvamento do quadril em paralisia cerebral: revisão sistemática

RESUMO

O desequilíbrio e a espasticidade muscular, associados à coxa valga e à anteversão femoral persistente, comprometem o desenvolvimento do quadril na paralisia cerebral e podem resultar em dor crônica e até luxação. Alguns desses quadris são submetidos a cirurgias de salvamento decorrentes do grave impacto das suas alterações na qualidade de vida. Fizemos uma revisão sistemática da literatura para comparar os resultados das principais técnicas aplicadas para salvamento do quadril nesses indivíduos. A busca na literatura teve como

Palavras-chave:

Paralisia cerebral

Salvamento

Quadril

Fêmur

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<http://dx.doi.org/10.1016/j.rboe.2015.06.003>

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foco estudos que avaliaram resultados de cirurgias de salvamento do quadril em paralisia cerebral, publicados de 1970 a 2011, presentes nas bases de dados Embase, Medline, Pubmed, Scielo e Cochrane Library. Apesar de os resultados obtidos não serem estatisticamente comparáveis, essa revisão sistemática demonstra que as cirurgias de salvamento do quadril devem ser indicadas após avaliação individual de cada paciente, decorrente do amplo espectro de apresentações da paralisia cerebral. Logo, aparentemente, não há uma técnica cirúrgica superior às outras, mas sim indicações diferentes.

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Introduction

Hip development is determined through the constant actions of the musculature on this joint. Individuals with cerebral palsy (CP) are born with normal hips.¹ However, through the muscle imbalance generated by the spasticity present in this condition, in association with coxa valga and uncorrected femoral anteversion, hip development becomes compromised and this results in deformities of different levels of severity.^{1,2}

The incidence of hip subluxation and dislocation in spastic CP cases is highly variable, between 7% and 60%. Higher prevalence, from 33% to 70%, is found among patients with greater neurological impairment, quadriplegic cases or cases presenting GMFCS (Gross Motor Function Classification System) levels III, IV or V.^{3,4} Pain, difficulty in sitting down, pressure ulcers and impaired perineal hygiene are the main problems resulting from these alterations when the condition goes untreated.⁵

The aim of the treatments is to obtain a pain-free hip that allows adequate range of motion and provides better positioning and capacity to sit, in addition to implementing hygiene and general care measures.^{6,7} The results from the various closed or surgical treatments are directly linked to the qualities of the joint surface, acetabular cartilage and femoral head.⁸ Thus, younger patients benefit more from closed reconstruction procedures. On the other hand, hips that present chronic subluxation or dislocation may require salvage surgery, such as femoral head resection, arthroplasty, osteotomy or arthrodesis.^{9,10}

The literature on this subject is sparse, particularly with regard to presenting groups of patients that can be compared. There are no controlled prospective studies, systematic reviews or meta-analyses comparing the various hip salvage surgery techniques applied in CP cases. The objectives of the present study were to carry out a systematic review of the literature in order to compare the results from the main techniques applied for hip salvage among these individuals and answer the following question: which surgical technique is the most effective for treating dislocated and painful hips in CP cases, given the salvage surgery methods available?

Material and methods

A qualitative and quantitative systematic review of the literature was carried out, which had the aim of identifying all the evidence on hip salvage surgery in CP cases.

Two reviewers separately performed a literature search focusing on studies that evaluated the results from hip salvage surgery in CP cases, published between 1970 and 2012, using the Embase, Medline, PubMed, SciELO and Cochrane Library databases. For this, the following descriptors were used: *cerebral palsy, hip, femur, femoral* and *salvage*.

The studies selected complied with the inclusion and exclusion criteria that we established. All the inclusion criteria had to be met for the study to be used in the review. Prospective clinical trials, with or without randomization, and also retrospective studies, published in Portuguese, English, French, Spanish, German or Italian were included. Samples with at least 10 participants and length of follow-up of at least 12 months were also included.

The exclusion criteria were: case reports, editorials, descriptions of surgical techniques, studies that contained a sample of patients with neuromuscular diseases other than CP, and samples with mean age greater than 30 years ([Appendix A](#): protocol model).

The data-gathering process and inclusion and exclusion of studies were performed separately by the authors twice. The efficacy of each study was evaluated regarding improvement of pain, ease of personal hygiene, functional improvement, satisfaction and presence or absence of heterotopic ossification. The results were then evaluated and, lastly, a balanced and impartial synthesis was elaborated with due remarks relating to flaws in the evidence.

Results

Forty-eight published papers evaluating patients with CP and painful hips who underwent salvage surgery were found. Ten observational retrospective cohort studies were selected, while 38 studies were excluded (two descriptions of surgical techniques and 36 observational studies). No randomized and controlled experimental studies were found. Only one article presented a control group, and this study compared the techniques proposed by Castle and by McHale.¹¹

The surgical techniques used and analyzed in these selected articles were: hip arthroplasty ($n=1$)¹²; Schanz femoral valgus osteotomy ($n=1$)¹³; hip arthrodesis ($n=1$)¹⁴; McHale valgus osteotomy ($n=2$)^{15,16} and resection of the proximal end of the femur ($n=6$)^{2,16-19}. One study¹¹ compared two surgical techniques (resection of the proximal end of the femur and McHale valgus osteotomy) ([Table 1](#)).

The main reason for excluding the remaining studies was the surgical techniques used ($n=13$), which did not consist of

Table 1 – Characteristics according to surgical technique applied.

Surgical technique	Mean follow-up (months)	Sample size	Mean age (years)
Arthroplasty	57	11	17
Arthrodesis	63	14	15.4
Castle	53.9	94	17.6
McHale	54.5	21	10.1
Schanz	98	35	13.5
Total	65.28	175	14.72

Table 2 – Primary reasons for study exclusion.

Primary reason for exclusion	Number of studies
Surgical technique	13
Language	6
Heterogeneous patient sample	7
Mean age > 30 years	53
Sample size	
Description of surgical techniques	2
Poorly described results	2
Total	38

hip salvage surgery, but rather, were interventions for improving the position of the hip through releasing soft tissues, along with acetabular and femoral osteotomies. Other causes for exclusion were, in decreasing order: heterogeneous samples ($n=7$), language ($n=6$), mean age greater than 30 years ($n=5$), inadequate sample size ($n=3$), description of surgical techniques ($n=2$) and poorly described results ($n=2$) (Table 2).

The studies selected were published between 1999 and 2009. The sample size ranged from 7 to 35 patients, and the summed total was 175 individuals. The mean age of the patients in the included studies was 14.72 years. The older patients underwent resection of the proximal end of the femur (mean of 17.6 years), while the younger ones underwent the McHale valgus osteotomy technique (mean of 10.1 years). The follow-up of all the series was greater than 21 months, with

a mean total of 65.28 months. These were all observational cohort studies, with level IV evidence (Table 3).

Discussion

There is great debate in the literature concerning surgical treatment for painful hips in CP cases, especially in relation to those with some level of dislocation, because there is no consensus regarding whether this factor is what causes the painful condition.¹⁰ Treatment decisions need to be based on careful individualized clinical observations on each patient.^{14,20}

This review of the literature consisted of qualitative analysis on the results from the selected studies. Quantitative analysis, i.e. meta-analysis, was not possible because of the heterogeneity of the variables evaluated and the interpretation of the outcomes obtained from the studies, which therefore made the results not statistically comparable.

In many situations, there are no controlled randomized studies in the literature, and only data from observational studies are available. Although the initial preference for systematic reviews is to restrict them to randomized prospective studies, the number of reviews and meta-analyses published involving observational studies has increased substantially over the past four decades, with the aim of filling this gap.²¹

The main obstacle relating to reviews with observational studies is the impossibility of controlling the sample and the variables evaluated because of their retrospective nature. Therefore, it is not always possible to compare the selected studies through meta-analysis.²¹

Total and partial hip arthroplasty

Only one study evaluated the use of partial arthroplasty as a hip salvage technique in CP cases.¹² Gabos et al.¹² used the humeral component of a shoulder prosthesis after resection of the proximal end of the femur. Over a period of four years and nine months, 11 patients of mean age 17 years who presented quadriplegic cerebral palsy and were unable to walk were evaluated. Pain criteria and improvement of perineal hygiene were evaluated by means of questionnaires applied over the telephone. In only one case was the evolution unfavorable,

Table 3 – Characteristics of the samples included.

Main author	Year of publication	Mean follow-up (months)	Sample size	Mean age (years)	Surgical technique used	Level of evidence
Gabos	1999	57	11p	17	Arthroplasty	4
Widman	1999	88.8	13p/18h	26.6	Castle	4
Fucs	2003	63	14p/14h	15.4	Arthrodesis	4
Ramos	2004	21	14p/14h	12.85	Castle	4
Leet	2004	40	7p/8p	19.9/14.5	Castle/McHale	4
Abu-Rajab	2007	40	15p/21h	16.2	Castle	4
Muthusamy	2008	92	25p/30h	15.5	Castle	4
Schejbalova	2008	98	35p/55h	13.5	Schanz	4
Van Riet	2009	69	13p/17h	5.75	McHale	4
Knaus	2009	42	20p/27h	15	Castle	4

p = patients; h = hips.

and this patient continued to have hygiene difficulties and hip pain.

Total hip arthroplasty in cerebral palsy cases is frequently recommended for older, skeletally mature patients.²²⁻²⁶ These individuals, given their greater age, mostly present less severe conditions with regard to spasticity and cognitive and motor deficit. This observation corroborates the finding that patients whose conditions are more severe cases die earlier.⁸ Thus, the success of total hip arthroplasty seems to be directly proportional to the severity of the clinical condition of CP, among other factors.

Arthrodesis

Only one of the studies used hip arthrodesis as an option for salvage surgery.¹⁴ Fucs et al.¹⁴ evaluated the evolution (mean follow-up of five years and three months) of 14 patients who underwent unilateral arthrodesis. This sample was composed of four diplegic individuals, eight quadriplegic patients and two with a mixed pattern (diplegia and dyskinesia), with a mean age of 15 years and five months. Functionally, four were able to walk (one of them only at home), three were only able to sit up and seven were permanently bedridden. All of these individuals presented complaints of severe unilateral hip pain.

The postoperative functional evaluation demonstrated absence of pain and better positioning (flexion-abduction) among all the individuals. Among the bedridden patients, five became able to remain seated and two started to walk. Despite the results obtained, the limits and contraindications of this technique include situations of the contralateral hip at risk or already compromised, and also the presence of spinal deformity.

Valgus proximal osteotomy of the femur

Regarding valgus proximal osteotomy of the femur, two studies were included. In one, Schanz osteotomy was applied,^{13,27} while in the other, the technique described by McHale was used.^{15,28}

These patients presented spastic quadriplegic CP, and their mean ages were 13.5 and 21 years, in the groups that underwent the McHale and Schanz surgeries, respectively. The mean minimum follow-up was 69 months.¹⁵ Among the criteria evaluated, both of these authors described improvements of pain in over 80% of their patients, although the painful condition persisted in 7.3%¹³ and 15.3%.¹⁵

Both of these techniques resulted in improvement of hip function and allowed better hygiene care, although the gain of mobility was only quantified (in degrees) in the group that underwent the McHale surgery: 90° of flexion or more was achieved by 76.9% of the patients and at least 35° of abduction in 69.2%. There were some complications such as heterotopic ossification, implant failure, postoperative infection and persistence of pain, but these occurred in a minimal number of cases.^{13,15}

Schejbalova et al.¹³ justified the choice of their procedure (Schantz valgus osteotomy) as being less invasive in relation to other surgical methods for the proximal femur, and stated that this should be applied to older children with dislocated hips for whom reconstructive surgery is not recommended.

Therefore, this method is an option for patients with severe quadriplegia, in relation to the technique of resection of the proximal end of the femur.

Resection of the proximal end of the femur

The most widely disseminated resection technique for the proximal end of the femur that is used as hip salvage surgery in CP cases was described by Castle and Schneider in 1978.²⁹ This method consists of interposition arthroplasty, i.e. subtrochanteric osteotomy, and involves removal of the femoral neck and head (which have been compromised by cartilaginous degeneration) and interposition of the musculature of the quadriceps between the stump and the acetabulum.

There are divergences regarding the postoperative management of patients who undergo the Castle surgery. The measures adopted have sought mainly to avoid ascension of the femoral stump and, therefore, recurrence of the painful condition.²⁹ To achieve this, methods such as cutaneous traction, skeletal traction, external fixators and pelvic immobilization with plaster casts have been used, depending on the author. The time of use of these methods has usually not been mentioned.^{11,27,29}

Six studies that used this technique, with its variations, such as treatment of the painful dislocated hip at different levels, were selected for this systematic review.^{2,11,16-19} Most of the samples were composed of patients with tetraparetic spastic CP who were unable to work, as seen in the study by Abu-Rajab et al.,¹⁸ in which all the individuals were classified at GMFCS level V.

The patients with CP who underwent resection of the proximal end of the femur were younger than those that underwent total hip arthroplasty. Their mean ages were 17.6 years and 38.1 years, respectively. These individuals were similar regarding clinical condition to those who underwent valgus osteotomy, and both groups once again differed from patients who undergo total hip arthroplasty, who are frequently able to walk and have better functional levels.

The postoperative results obtained with this technique were satisfactory in all the studies selected. What most differentiated the studies were the evaluation methods for pre- and postoperative pain, given that this is a subjective criterion and many of these patients presented cognitive deficit. Widman et al.¹⁶ used the quantity of analgesic pills that the patient was consuming per day. Another author measured the evolution of pain by means of an analog scale (1-10), which was applied over the telephone.¹¹

Regarding function and perineal hygiene, this surgical treatment was also shown to lead to an improvement in range of motion and consequently in perineal care. Evaluation of perineal hygiene was found in all the studies, with improvements of 62%¹⁶ to 100%,^{17,18} from assessment of the ease of perineal cleaning after the procedure.¹⁶⁻¹⁹ In turn, the evolution of the range of motion was assessed in most studies through measurement and comparison of abduction amplitudes before and after the operation.^{16,17} All the studies presented gains in range of motion.

The complications inherent to this procedure include ascension of the femoral stump, return of pain or absence of improvement and heterotopic ossification. Only one author

recommended radiotherapy (RT), with 700 cGy, to prevent heterotopic ossification. In that study, using Brooker's classification, means of 2.7 among patients without RT and 0.8 among those who underwent RT were obtained.¹⁶

Finally, the only comparative study on hip salvage surgery techniques was conducted by Leet et al.¹¹ in 2004. The evolution (mean follow-up of 3.4 years) of 15 individuals who had undergone the Castle and Schneider²⁹ (n=7) and McHale et al.²⁸ (n=8) techniques were compared by means of a questionnaire applied over the telephone, to assess satisfaction, mobility, perineal hygiene and pain, on a scale from 0 to 10. The group that underwent the Castle and Schneider technique presented a preoperative score of 8.2, which decreased to 2.9 after surgery. The group that underwent the McHale technique had a preoperative score of 8.0, which decreased to 4.8 after surgery. The satisfaction score among the individuals who underwent the Castle and Schneider technique was 9, while those who underwent the McHale technique had a score of 7.7. The differences obtained in both techniques were not statistically significant.

Final remarks

Although the results obtained were not statistically comparable, this systematic review demonstrated that hip salvage surgery should be indicated after individual assessment of each patient, because of the broad spectrum of presentation of CP. Thus, there does not seem to be any surgical technique that is superior to any other: rather, there are different indications.

Conflicts of interest

The authors declare no conflicts of interest.

Anexo 1. Protocol for inclusion/exclusion of studies

- 1- Surgical technique:
- 2- Sample size (n):
- Male sex:female sex:
- 3- Mean length of follow-up:
- 4- Mean age:
- 5- Evolution criteria: () pain () hygiene () mobility () satisfaction () heterotopic ossification () others:_____

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