

# Long-Term Results and Outcome Predictors in One-Stage Hip Reconstruction in Children with Cerebral Palsy

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**Background:** One-stage hip reconstruction is the gold standard for treatment of hip displacement in children with cerebral palsy. The aims of this study were (1) to report the subjective clinical, objective clinical, and radiographic outcomes; and (2) to investigate outcome predictors, including the influence of the following risk factors: femoral head shape, migration percentage, direction of migration, and age at surgery.

**Methods:** We reviewed 168 hip reconstructions (eighty-two right, eighty-six left) in 121 patients (101 male, twenty female) at a mean follow-up of  $7.3 \pm 4.6$  years (range, four to eighteen years). Surgical outcomes were assessed on the basis of the pain intensity and frequency (measured on 10-point visual analog scales) as well as GMFCS (Gross Motor Function Classification System) and MCPHCS (Melbourne Cerebral Palsy Hip Classification System) scores and postoperative migration percentage. The effects of femoral head shape, preoperative migration percentage, direction of migration, and age at surgery on surgical outcome were assessed by multivariate regression adjusting for potential confounders including sex, triradiate cartilage status, type of cerebral palsy, and surgical technique.

**Results:** Pain intensity and frequency were reduced significantly. Preoperative femoral head shape had no significant effect on the changes in pain, MCPHCS grade, and GMFCS level. The preoperative migration percentage was the most influential risk factor with respect to postoperative outcome. Age at surgery had no effect on the changes in pain score and GMFCS level. The overall surgical complication rate was 10.5%.

**Conclusions:** Our data on 168 hip reconstructions at a mean follow-up of seven years showed significant and clinically meaningful improvements in pain intensity and frequency as well as in clinical scores and hip coverage. Analysis of potential risk factors showed only the preoperative migration percentage to have a relevant influence on outcomes.

**Level of Evidence:** Therapeutic Level IV. See Instructions for Authors for a complete description of levels of evidence.

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Cerebral palsy (CP) is the leading cause of physical disability during childhood<sup>1</sup>. The CDC (Centers for Disease Control and Prevention) reports a prevalence of 1.5 to 4 per 1000 births, and improved neonatal and obstetric care have resulted in decreased mortality of preterm infants at

the expense of a further increase in CP cases. Children with CP have health-care costs twenty-six times higher than those of “typically developing children,” amounting to an approximately \$1 million lifetime cost of care, according to a 2003 study (<http://www.cdc.gov/mmwr/preview/mmwrhtml/mm5303a4.htm>).

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This makes CP an important issue not only for pediatric orthopaedic surgeons but also for health care in general.

Only one-half of children with CP will learn to walk independently, and one-third will achieve only severely limited or no walking ability. The hip plays a major role in walking ability and motor function, and up to 75% of children with CP develop hip displacement<sup>2-6</sup>. This percentage is directly related to the GMFCS (Gross Motor Function Classification System) level<sup>7</sup>. The GMFCS allows defining categories of mobility for these patients and is directly related to the extent of hip displacement<sup>7</sup> (see Appendix).

In response to these facts, orthopaedic treatment for hip dislocation in CP aims at screening<sup>3,5,8</sup>, early detection<sup>2,4</sup>, and timely surgical correction at still-reversible stages<sup>9-11</sup>. Temporizing procedures such as botulinum toxin<sup>12</sup> or soft-tissue lengthening are available<sup>13,14</sup>; however, definitive treatment typically includes pelvic and/or femoral osteotomies as unilateral or bilateral procedures<sup>15-17</sup>.

Good subjective as well as objective short and intermediate-term results have been reported for acetabuloplasty and/or femoral, derotational, shortening, and varus-producing osteotomy in a number of cohort studies with small to medium sample sizes<sup>11,15,18-21</sup>. However, to the best of our knowledge, no large-scale, long-term study on the subject of hip reconstruction in CP has been published.

The objectives of this study were twofold. First, we wished to report the subjective clinical, objective clinical, and radiographic outcomes of the surgical reconstruction of 168 hips in patients with CP at a mean follow-up of seven years. Second, we wished to study the significance and magnitude of the influence of the above-mentioned risk factors on postoperative outcomes, including adjustment for potentially confounding covariates such as sex, triradiate cartilage status, type of CP, and surgical technique.

## Materials and Methods

Ethical approval for this study was granted under the audit provisions of our local institutional review board.

### Patients

All data from 239 patients (GMFCS levels I through V) with a migration percentage (MP) of >30% who underwent reconstructive surgery including pelvic and femoral correction were collected prospectively and continuously in a database since 1989. The data were analyzed retrospectively, and patients with complete data on all studied variables were included in this analysis. A total of 118 patients were excluded because of missing data or attrition. (Four died, thirty-six had missing radiographs, fourteen had incomplete clinical data, and sixty-four were not available for follow-up.) There were no significant differences between the included and excluded patients in terms of age at surgery ( $p = 0.5903$ ), preoperative GMFCS level ( $p = 0.2830$ ), pain intensity ( $p = 0.7571$ ), or pain frequency ( $p = 0.6873$ ). However, fewer patients were female in the included group (17%) than in the excluded group (29%,  $p = 0.009$ ). Complete data sets were available for 168 hip reconstructions (eighty-two right and eighty-six left) performed in 121 patients (101 male and twenty female) with confirmed acetabular dysplasia. Of the included patients, 119 (98%) were classified as having quadriplegia and two (2%), as having diplegia. The preoperative GMFCS level was classified as V in eighty-one patients (67%), IV in twenty-nine (24%), III in four (3%), and II in the remaining seven (6%). One hundred and one patients (83%) cited pain as the primary reason to undergo surgery, and the remaining twenty (17%) cited function (i.e., limited walking,

TABLE I Patient Demographics\*

Mean age (range) (yr)	11 (4 to 23)
Sex	101 M, 20 F
Side	82 R, 86 L
Topographical distribution	119 quadriplegic, 2 diplegic
Mean follow-up (range) (yr)	7.3 (4 to 18)
Median GMFCS level (IQR)	V (IV to V)
Pelvic osteotomy	168 Pemberton
Femoral osteotomy	113 shortening VDO, 8 VDO

\*VDO = varus derotation osteotomy.

standing, or sitting ability). The mean age at surgery (and standard deviation) was  $11.3 \pm 3.7$  years (range, four to twenty-three years). The mean duration of follow-up at the time of analysis was  $7.3 \pm 4.6$  years (range, four to eighteen years) (Table I).

### Surgical Procedure

All surgical procedures were performed under general anesthesia with antibiotic prophylaxis. Preoperative planning was done by hand with use of Dunn view radiographs, true anteroposterior radiographs with internal rotation of the hip, and three-dimensional CT (computed tomography).

All patients were treated with a modified Pemberton osteotomy<sup>16,17,22</sup> with a varus derotational osteotomy or shortening varus derotation osteotomy<sup>9,10</sup>. The femoral osteotomy was secured with an AO blade plate, and the goal was a neck shaft angle of  $120^\circ$  to  $125^\circ$  with  $20^\circ$  to  $25^\circ$  of antetorsion. The acetabuloplasty was secured with 0.062-in (1.6-mm) Kirschner wires if necessary. Open reduction was performed if necessary (as indicated by intraoperative arthrography). Soft-tissue procedures including release, transfer, or lengthening of the psoas, rectus, and adductors were added as appropriate<sup>13,14,23-25</sup>. If bilateral procedures were indicated, they were performed during the same surgical session rather than in a staged fashion. The hip reconstruction procedure has been described in detail by the senior author and a colleague<sup>26</sup>. All surgical steps were included as ordinal variables in the multivariate statistical model.

Postoperatively, pain and spasticity were treated with peridural anesthesia and diazepam (0.1 to 0.2 mg/kg every six to eight hours). A spica cast was applied, and this was replaced by an abduction device after six weeks (for patients treated through 2003) or after two to three weeks (after 2003). The spica cast is used for pain management by release of muscle tension, and it is not meant to maintain or improve reduction. Postoperative visits included standardized radiographs and physical examination and were performed at six, twelve, and twenty-four weeks and yearly thereafter. Hardware removal was performed more than six months after the initial procedure.

### Outcome Measures

Three types of end points—subjective clinical, objective clinical, and radiographic—were utilized. The subjective clinical end points were pain intensity and pain frequency, measured on a 10-point VAS (visual analog scale) anchored at “no pain” and “worst pain imaginable” for intensity and at “no pain” and “permanent pain” for frequency. Pain was assessed routinely during all visits prior to and after surgery. In this population, 34% of the children were uncommunicative, and their pain levels were obtained from parents or caregivers; 66% were able to express their pain levels.

The objective clinical end point was the GMFCS<sup>27-29</sup>. All children who were treated prior to the publication of the GMFCS in 1997 were classified on the basis of the existing data.

Radiographic assessment was performed with use of the MP<sup>30</sup> and the revised version of the MCPHCS (Melbourne Cerebral Palsy Hip Classification

TABLE II Outcome Measures

	Preoperative	Final Follow-up	P Value
Pain intensity*	6.9 ± 1.9	1.5 ± 2.4	<0.001
Pain frequency*	5.7 ± 1.7	1.4 ± 1.1	<0.001
GMFCS level†	V (IV to V)	V (IV to V)	<0.001
MCPHCS grade†	4 (4 to 5)	1 (1 to 3)	<0.001
MP* (%)	77 ± 23	10 ± 16	<0.001

\*The values are given as the mean and the standard deviation.

†The values are given as the median, with the IQR in parentheses.

System)<sup>31</sup>. The MCPHCS is a radiographic classification system that includes joint congruency and alignment as well as acetabular and femoral head deformity (see Appendix). The MP describes the subluxation of the femoral head out of the acetabulum relative to the width of the head<sup>32</sup> (see Appendix). Perioperative and postoperative complications are reported descriptively.

The outcome measures utilized in this study have been tested for validity and reliability in prior studies<sup>33</sup>.

### Potential Outcome Predictors

The first risk factor in the analysis was femoral head shape, classified as A through D. Type A is a normally configured head with intact cartilage. Type B has medial flattening and a head-in-neck deformity. Type C is a progression from type B in which a lateral trough is also present. Type D is consistent with the complete loss of the round shape<sup>30,34</sup> (see Appendix).

The second risk factor was the preoperative MP<sup>30</sup> (see Appendix). An MP of ≥30% but <100% is defined as hip displacement, and an MP of ≥100% is considered hip dislocation<sup>6</sup>.

The third risk factor was the direction of migration as assessed by three-dimensional CT. A clock face was projected on the plane of the acetabulum to identify this direction. The clock face was divided into eight segments, the top five of which were used. These segments were designated as anterior, antero-lateral, straight lateral, posterolateral, and posterior<sup>35</sup> (see Appendix).

The fourth risk factor in the analysis was age at surgery. Inclusion of this factor was based on earlier published data indicating that the rate at which correction is lost after hip reconstruction is greater in younger patients<sup>4,5</sup>.

### Potential Confounders

Numerous potential confounders exist in such a large and diverse population. Although we attempted to avoid confounding bias by using strict inclusion and exclusion criteria, we also tested for the effect of certain potential confounders. Specifically, we included details of the surgical technique (psoas release or transfer, rectus release or transfer, and open or closed reduction), triradiate cartilage status, and sex in our statistical analysis.

### Statistical Analysis

The database was checked for completeness and consistency before the analysis. Unit nonresponse (absence of all data for a patient) was dealt with by case deletion. Data for complications were retained even in case of deletion. Data were tested for normality with use of the Kolmogorov-Smirnov test.

Postoperative outcomes are reported as the mean and 95% CI (confidence interval) or as the median and IQR (interquartile range). The Mann-Whitney U test was used for comparisons involving ordinal data, and the t test was used for other comparisons.

TABLE III Results of Multivariate Analyses\*

Effect of	Effect on	Coefficient†	95% CI	P Value
Head shape	Pain intensity	-0.29	-0.75 to 0.15	0.193
	Pain frequency	-0.25	-0.64 to 0.15	0.221
	GMFCS	-0.09	-0.18 to 0.01	0.072
	MCPHCS	-0.07	-0.30 to 0.17	0.572
	MP	0.03	-0.02 to 0.08	0.214
Preop. MP	Pain intensity	1.68	0.07 to 3.29	0.041†
	Pain frequency	-0.48	-1.91 to 0.95	0.512
	GMFCS	0.01	-0.33 to 0.35	0.969
	MCPHCS	1.55	0.76 to 2.35	<0.001†
	MP	0.86	0.75 to 0.96	<0.001†
Direction of migration	Pain intensity	0.3	-0.19 to 0.79	0.234
	Pain frequency	0.04	-0.40 to 0.49	0.844
	GMFCS	-0.02	-0.12 to 0.08	0.655
	MCPHCS	0.31	-0.07 to 0.55	0.111
	MP	0.06	0.01 to 0.11	0.015†
Age at surgery	Pain intensity	-0.004	-0.013 to 0.004	0.277
	Pain frequency	-0.005	-0.010 to 0.002	0.192
	GMFCS	-0.0001	-0.002 to 0.002	0.874
	MCPHCS	-0.001	-0.010 to -0.001	0.008†
	MP	-0.0003	-0.002 to 0.001	0.533

\*Adjusted for confounders (see text). †Significant effect.

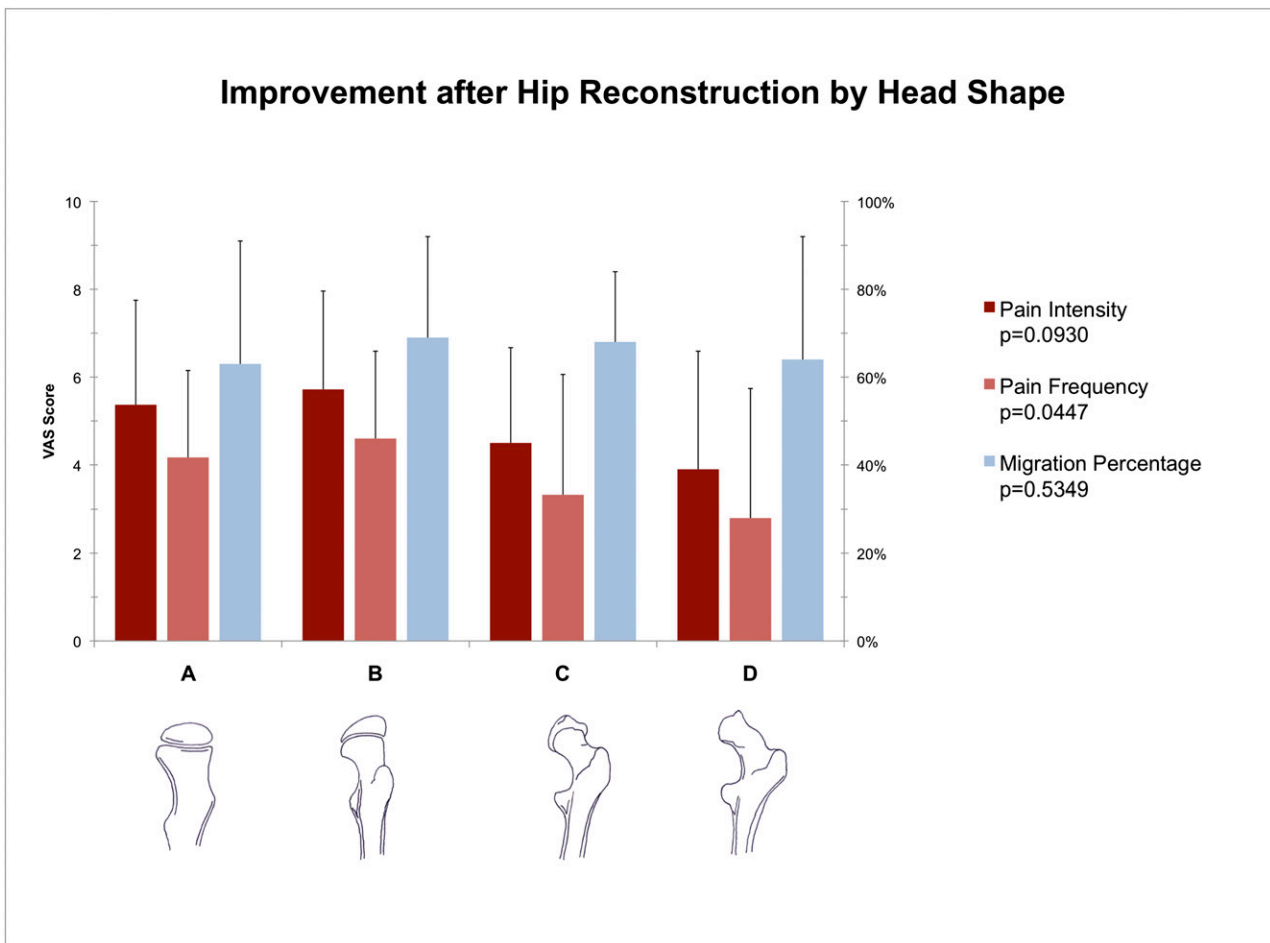


Fig. 1 Improvement in pain scores and MP following hip reconstruction according to the preoperative femoral head shape. The error bars indicate the standard deviation. Values adjusted for potential confounders are given in the text.

The significance and magnitude of the influence of the four risk factors of head shape, age, MP, and direction of migration were tested by means of multivariate, backward stepwise regression modeling. Head shape and direction of migration were treated as ordinal variables and the others, as continuous variables. The potential cofounders outlined above were used as covariates. The cutoff for significance was defined as  $p = 0.1$  rather than  $p = 0.05$  to account for the low power of such modeling. The fit of the final regression model was tested by assessing the normality of the distribution and the homoscedasticity of the residuals.

All statistical analyses were performed on an intention-to-treat basis with use of Stata (version 12; StataCorp, College Station, Texas) by an epidemiologist with advanced training in biostatistics. A  $p$  value of 0.05 was considered significant in all tests except the backward stepwise modeling. Holm-Bonferroni adjustment was used to account for multiple testing.

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No external funding was obtained for this study.

## Results

### Outcome After Hip Reconstruction

There was a significant improvement in pain, with the mean intensity differing by 5.4 points (95% CI, 5.1 to 5.8) and the mean frequency differing by 4.3 points (95% CI, 3.9 to 4.6 points). There was also a small but significant improvement in

the GMFCS level, with the mean differing by 0.2 point (95% CI, 0.1 to 0.3 point). The mean MCPHCS grade differed by 2.6 (95% CI, 2.4 to 2.8). The MP improved by 66% (95% CI, 62% to 70%) (Table II).

### Risk Factors in the Surgical Outcome

In the multivariate regression model (Table III), preoperative head shape had no significant effect on the changes in pain intensity ( $p = 0.193$ ), pain frequency ( $p = 0.221$ ), GMFCS level ( $p = 0.072$ ), MCPHCS grade ( $p = 0.572$ ), or MP improvement ( $p = 0.214$ ) (Fig. 1).

Unsurprisingly, the preoperative MP was the most influential risk factor affecting the postoperative outcome. The MP showed a significant association ( $p = 0.041$ ) with pain intensity, with a mean increase in pain of 1.7 points (95% CI, 0.07 to 3.3 points) per 1% increase in MP, but no significant association with pain frequency ( $p = 0.512$ ). The preoperative MP had no effect on the GMFCS improvement ( $p = 0.969$ ), but it did have a significant effect on the improvement in the MCPHCS ( $p < 0.001$ ), which is plausible since femoral head subluxation or dislocation is a part of the latter hip classification system.

The direction of femoral head displacement had no effect on the changes in pain intensity ( $p = 0.234$ ) and frequency ( $p = 0.844$ ). Our data do not support an association between the GMFCS level ( $p = 0.655$ ) and the direction of migration or between the MCPHCS grade ( $p = 0.111$ ) and the direction of migration. Unsurprisingly, there was a significant association between the direction of migration and the MP correction ( $p = 0.015$ ). However, when the five directions were analyzed separately, this association was found to be based on the influence of lateral migration ( $p = 0.001$ ); the other directions showed no significant influence on MP improvement. This is especially surprising considering the difficulties in treating pure anterior or posterior dislocations.

Age at surgery had no effect on the changes in pain intensity ( $p = 0.277$ ), pain frequency ( $p = 0.192$ ), and GMFCS level ( $p = 0.874$ ). However, age at surgery did show a significant negative association with the change in the MCPHCS grade ( $p = 0.008$ ). Briefly, each one-year decrease in patient age at surgery corresponded to a 6.3% decrease in the MCPHCS improvement at the final follow-up. The MP showed no association with age at surgery ( $p = 0.533$ ). Also, we found no evidence for an association between age and femoral head shape.

### Complications

The overall surgical complication rate was 10.5% per treated hip. There were no neurovascular injuries. There was one case (0.6%) of septic arthritis, which required surgical revision and debridement; the patient responded well to this treatment and maintained good hip alignment.

Recurrent hip dislocations requiring repeat hip reconstruction occurred in two of the 168 hips. Eleven hips (6.6%) showed initial radiographic progression of the femoral head deformity, with subsequent delayed remodeling that took up to two years to complete. There was one case (0.6%) of acetabular necrosis that resolved with conservative treatment. There was one case of heterotopic ossification (Brooker grade II)<sup>36</sup>. One patient required revision because of excessive derotation leading to femoral retrotorsion. Three patients (2.5%) had a supracondylar femoral shaft fracture and underwent open reduction and internal fixation with plates. Thirty-six patients (29.8%) had a medical complication, including a chest infection in 13.2% (sixteen), aspiration in 1.7% (two), decubitus ulceration (involving the heel or coccyx) in 9.9% (twelve), deterioration of a seizure disorder in 2.5% (three), and urinary tract infection in 2.5% (three).

### Discussion

This study provides evidence that surgical hip reconstruction produces substantial and lasting reduction of pain intensity and frequency in children with CP. Interestingly, we found only limited evidence for a detrimental influence of four selected risk factors on surgical outcomes, suggesting that hip reconstruction with attention to detail will produce beneficial results in most patients.

This study has some limitations; the most obvious is the level of attrition. However, given the duration of follow-up and

the complexity of maintaining contact with these children and adolescents, we consider the follow-up percentage not particularly poor and in keeping with the existing literature<sup>37</sup>. Also, the outcome assessment instruments used are not as well developed as some of the currently available instruments, but all data were consistently recorded on the same scales throughout the twenty-five-year study duration (1989 to 2014). Furthermore, our surgical techniques were somewhat heterogeneous. For example, approximately half of the patients received a psoas transfer or release. We compared the outcomes according to surgical technique but found no differences.

Our data showed that hip reconstruction leads to consistent and predictable improvement in pain frequency and intensity at a mean follow-up of seven years. The level of pain reduction was significant but also clinically substantial, leading to a level consistent with the “normal” pain experience of a healthy child during the course of childhood injuries and sickness<sup>38</sup>. The improvements in the GMFCS and MCPHCS functional scores were also significant, although the GMFCS improvement was only small in magnitude. The small change is not surprising, as many factors are included in these compound measures, even if the hip is among the predominant ones.

Factors influencing the outcome were of further interest. Our results clearly showed that head shape had no significant effect on changes in pain intensity and pain frequency. Thus, femoral head deformity is not a contraindication for reconstructive surgery. A prior study by Castle and Schneider indicated better outcomes and fewer complications after resection arthroplasty in patients who had a deformed femoral head<sup>39</sup>. A recent systematic review<sup>34</sup> that included newer surgical techniques<sup>39</sup> indicated that the current literature does provide evidence to favor head-preserving treatment over resection arthroplasty. This corroborates our data, showing that even in a hip with a badly deformed femoral head, reconstruction can be a very successful procedure, leading to both pain reduction and functional improvements.

The preoperative MP was the most influential risk factor affecting the postoperative outcome. The MP had a highly significant association with pain intensity, with a mean increase in pain of 1.7 points per additional percent of migration. However, with a mean MP of almost 80%, the patients in our population had a fairly sizable hip extrusion. The influence of the preoperative MP on postoperative outcomes is most likely smaller in patients with a smaller MP. Likewise, our data suggest initiating hip reconstruction early, before the MP gets too large.

However, the MP is limited in cases of anterior-posterior migration. Three-dimensional imaging of the hip with CT<sup>40</sup> may be indicated in such patients<sup>32</sup>. Three-dimensional CT scanning was performed routinely at our institution prior to evaluating the preoperative situation for optimal positioning during surgery<sup>26</sup>. With use of this protocol, our data indicate that the direction of migration had little effect on the change in pain intensity or frequency.

Finally, with much improved survival, patients with CP are reaching older ages<sup>41</sup>. Recent studies have shown that 95%



of patients with CP now survive for more than thirty years<sup>42,43</sup>. Consequently, we wanted to include age as a risk factor for post-operative outcomes of hip reconstruction. However, age at surgery had no effect on the changes in pain intensity, pain frequency, and GMFCS level.


The rates of surgical (11%) as well as medical (21%) complications were not excessive or atypical for the extent of surgical intervention. We did observe three fractures, which might have resulted from use of a spica cast. However, we did see very positive results with the use of spica casting in terms of reduced pain postoperatively. In a study by Skaggs et al. in 1999, the refracture rate after femoral shaft external fixation in children was 12%, compared with the 0.02% overall fracture risk reported by Hinton et al. during the same year<sup>44,45</sup>. Our fracture rate of 2.5% seems comparable to that of Skaggs et al. Also, we cannot be absolutely certain that there were no additional cases of mild osteonecrosis that were missed because of spontaneous resolution or lack of clinical symptoms.

Prior research corroborates our findings, although either in smaller populations or with shorter follow-up. In 2013, Dhawale et al.<sup>18</sup> reviewed the long-term radiographic outcome, complications, and revision procedures in nineteen children with quadriplegic CP. At a mean follow-up of 11.7 years, the MCPHCS was grade 2 in sixteen hips, grade 3 in five, and grade 5 in one. Five complications were noted in four hips (21%)<sup>18</sup>. The conclusion was that good long-term outcomes can be achieved in preadolescent children with quadriplegic CP after reconstruction of the hip, but regular follow-up is required. Mallet et al. reported on a similarly small group of twenty patients with eight years of follow-up and showed good hip coverage, although clinical outcomes were not assessed<sup>37</sup>. One issue that is, however, still somewhat controversial is whether there is a necessity for bilateral procedures, especially in cases with pelvic obliquity, a large MP, or contralateral contractures<sup>15-17,46</sup>. There

is research to support such an approach; nevertheless, our current strategy is to address only those hips that show unequivocal subluxation, whether it is present unilaterally or bilaterally.

In conclusion, our data on 168 hip reconstructions with a mean follow-up of seven years showed significant and clinically meaningful improvements in pain intensity and frequency as well as in clinical scores and hip coverage. Of note, the severity of femoral head deformity did not influence outcomes, suggesting that reconstruction is no less successful in a hip with a gravely deformed head than in one with mild deformity.

## Appendix

 Figures illustrating the GMFCS, revised MCPHCS, MP, head shape classification, and migration direction classification are available with the online version of this article as a data supplement at [jbjs.org](http://jbjs.org). ■

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