Can Chiari Osteotomy Favorably Influence Long-term Hip Degradation in Multiple Epiphyseal Dysplasia and Pseudoachondroplasia?

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Background: Multiple epiphyseal dysplasia (MED) and pseudoachondroplasia (PSACH) are congenital skeletal disorders characterized by irregular epiphyses, mild or severe short stature and early-onset osteoarthritis which frequently affect the hips. The current study evaluates the long-term results of the Chiari osteotomy in MED and PSACH patients.

Methods: Twenty patients (14 MED and 6 PSACH) were retrospectively included. Clinical assessment used the Postel Merle d'Aubigné (PMA) score and the Hip disability and Osteoarthritis Outcome Score (HOOS). Risser index, Sharp angle, acetabular depth index, center-edge angle, Tönnis angle, and femoral head coverage were measured on the preoperative radiographs and at last follow-up. The Treble index, which identifies the hip at risk in MED patients, was also determined. Stulberg classification (grades I to V) was used to evaluate the risk of osteoarthritis in the mature hips. Statistical analyses determined differences between preoperative and postoperative data. The Kaplan Meier method was used to calculate the survival rate of the operated hips using total hip arthroplasty as the endpoint.

Results: Thirty-three hips which underwent a Chiari osteotomy were reviewed. The average follow-up was 20.1 years. The PMA scores were significantly better at last follow-up than preoperatively. All radiographic parameters significantly improved. Moreover, the Sharp angle, center-edge angle, and femoral head coverage improved to a normal value at hip maturity. All of the operated hips had a Treble index of type I. At hip maturity, a majority of hip were aspherical congruent (Stulberg grades of III and IV). The survival rate of the operated hips was 80.7% at 24 years postoperative.

Conclusions: The Chiari osteotomy is a satisfying solution for severe symptomatic hip lesions in MED and PSACH patients. At long-term follow-up, this procedure lessens pain and improves hip function, which delays total hip arthroplasty indication.

Level of Evidence: Level IV.

Key Words: multiple epiphyseal dysplasia, pseudoachondroplasia, hip dysplasia, chiari osteotomy, skeletal dysplasia, *COMP*-gene

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Multiple epiphyseal dysplasia (MED) and pseudoachondroplasia (PSACH) are congenital skeletal disorders that are part of the same bone dysplasia family.[1] PSACH is an autosomal dominant disorder and results exclusively from mutations in cartilage oligomeric matrix protein (COMP).[2,3] MED can be caused by mutations in 6 different genes, including *COMP*, and is inherited in an autosomal dominant pattern in 75% of cases.[2,4]

The clinical and radiologic disease spectrum is heterogeneous and ranges from irregular ossification of the epiphyses with joint pain and stiffness to short stature and deformities of the limbs.[4] The weight-bearing joints could be severely affected with the development of an early-onset osteoarthritis.[5,6] The epiphyseal involvement is similar from a biochemical point of view in MED and PSACH patients but PSACH usually results in a more severe pathology than MED. In PSACH patients, coxarthrosis appears early, at a median age of 22.5 years.[7,8] In MED patients, it was reported that 33% of the hips showed signs of osteoarthritis before the age of 40.[6]

There is currently no appropriate therapeutic management for affected hips during childhood and adolescence and no recommended preventive treatment for the development of osteoarthritis in the patients with MED or PSACH. Surgery was described for treatment of hip disorders in MED patients but only midterm results in heterogeneous studies are available.[9–11]

The Chiari osteotomy was first described in 1955.[12] This extra-articular procedure provides bony coverage with interposed hip joint capsule and medialization of the hip joint that contribute to improve biomechanics and hip stability.[13,14] The weight-bearing surface of the hip is increased in order to decrease the load on the joint surface.

Based on our experience, we believe that the Chiari osteotomy reduces pain in MED and PSACH patients and in turn delays total hip arthroplasty (THA). The purpose of this study is to evaluate the clinical and radiologic results of Chiari osteotomy for hip disorders in MED and PSACH patients.

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METHODS

Population

After institutional review board approval, the inclusion criteria were: MED or PSACH patients who underwent Chiari osteotomy since 1975 in our institution and who have a minimal follow-up of 5 years. Twenty patients were included (33 procedures) among 22 patients (36 procedures). Two patients with incomplete files were excluded. There were 14 MED and 6 PSACH patients—11 male and 9 female. Twelve MED patients had the severe form of the pathology, called the Fairbank dysplasia according to the old classification.[4] The remaining 2 MED patients had the Ribbing dysplasia which is more localized to the hips. They showed an avascular necrosis (AVN) of the femoral head in addition to the epiphysis anomalies. The average age at surgery was 13.7 years old (range, 6.5 to 26 y). There were 13 bilateral procedures with a minimum of 3 weeks between the 2 procedures. Five diagnoses (2 PA and 3 MED) were confirmed by genetic analyses. All these patients presented a mutation of the *COMP* gene. Regarding our indications of treatment, we propose the Chiari osteotomy only to symptomatic patients that show a severe form of hip dysplasia. More recently, we started to propose the triple pelvic osteotomy in asymptomatic MED patients younger than 10 years old who show a type I hip according to Treble et al.[5]

Surgical Technique

We followed the original technique[12] for the osteotomy but performed it using an iliofemoral "bikini" approach.[15] The procedure was performed under fluoroscopy magnification on normal table without traction. The first steps of the procedure are similar to those of a pelvic osteotomy: the dissection of the interval between the sartorius and the tensor fascia lata and the splitting of the iliac apophysis for subperiosteal exposure of the lateral and medial tables of the ilium. The reflected head of the rectus femoris tendon is released from the acetabular roof to allow for exposure of the articular capsule. We use a straight chisel under fluoroscopic control to perform the osteotomy with an ascendant angle of at least 10 degrees going medially. The osteotomy is curvilinear in the anteroposterior plane: while in contact with the capsule, it passes under the anteroinferior iliac spine anteriorly and under the sciatic spine posteriorly. The curvilinear osteotomy is an important step to avoid posterior displacement of the distal fragment and thus prevent sciatic nerve palsy. Once the osteotomy is completed with the chisel, the medialization of the hip is performed using longitudinal traction combined with an abduction of the hip, applying a counter-support to the greater trochanter if needed. If the displacement is insufficient, a chisel is carefully inserted in the osteotomy for leverage. The longitudinal traction is stopped once the displacement is sufficient to cover the femoral head. When an osteosynthesis is not used, such as in the original technique, the patient is placed in a spica cast with the hip in extension, in neutral rotation, and in a 30-degree abduction. We now favor using an osteosynthesis with Kirschner wires, which offers the advantages of avoiding a secondary displacement and a spica cast. The postoperative immobilization is then performed through skin traction for 3 to 6 weeks.

Preoperative Data

Preoperative data was retrospectively collected from the medical files.

Sequential radiographic studies of hip joints were evaluated for occurrences of femoral head AVN. It was defined as the sequential radiographic change of all or part of stages including sclerosis, subchondral crescent sign, resorption of the preexisting bone tissue, and reossification superimposed on the already abnormal dysplastic epiphyses.[16]

The skeletal maturity using the Risser index, Sharp angle, acetabular depth index, center-edge angle, Tönnis angle, and femoral head coverage were measured on the preoperative radiographs. The Treble index was determined in the MED patients on whose radiographs the epiphysis was clearly visible.[5] It is a radiographic prognostic index using Sharp angle, coverage of the femoral head, epiphyseal index, and epiphyseal fragmentation. The hips are separated in 2 groups: type I is at high risk of precocious osteoarthritis and type II evolves quite normally (Fig. 1).

FIGURE 1. Treble index. A, Type 1 hip. B, Type 2 hip.

TABLE 1. Scoring System for the Final Assessment of the Results

Category	Pain	Limp	Range of Motion	Sharp Angle (Deg.)	Center-edge Angle (Deg.)	Uncoverage (%)
Excellent	No	No	Normal	<43	>25	<10
Good	Improved or not deteriorated			≤45	≥20	<20
Fair	Not deteriorated			≤50	≥15	≤30
Poor	Deteriorated			>50	<15	>30

From Hosny et al.[16]

Postoperative Data

The orientation of the osteotomy and the medialization percentage of the acetabulum were assessed on the first postoperative radiographs.[17] The Risser index, Sharp angle, acetabular depth index, center-edge angle, Tönnis angle, and femoral head coverage were measured on radiographs at last follow-up.

Patients were asked to fill in the Hip disability and Osteoarthritis Outcome Score (HOOS) at last follow-up for each native hip operated with a Chiari osteotomy.[18] This score is widely used to evaluate the impairment caused by hip problems in the patients' daily activities. Their satisfaction about the results of the Chiari osteotomy was also recorded. The Postel Merle d'Aubigné (PMA) score[19] was determined by the physician.

The Stulberg grade[20] was determined on the radiographs at skeletal maturity. This classification describes the morphology of the femoral head and is correlated to a risk of precocious osteoarthritis. Grades I and II correspond to a congruent hip, grades III and IV to an aspherical congruent hip, and grade V to an aspherical incongruent hip. The final assessment of our results followed the method of De Waal Malefijt modified by Hosny et al[17] (Table 1). This score uses both clinical and radiologic results. We also reported the surgical revision including THA.

Statistical Analyses

The preoperative radiographic parameters and those at hip maturity were compared using paired the Student *t* test. This test was also used to determine the difference between preoperative PMA score and those at last follow-up. Potential correlations between age at surgery and the Stulberg grade and between final outcomes and the elapsed time since Chiari osteotomy were achieved. Differences were considered significant when the probability value (*P*) was <0.05.

The Kaplan-Meier method was used to determine the hips' survival rate after the Chiari osteotomy. The date of conversion to THA was considered as the end point.

RESULTS

The average age at last follow-up was 33.7 ± 13.6 y (range, 17 to 56 y). The average follow-up period was 20.1 years (range, 5.5 to 42 y). Figure 2 shows age distribution at the time of the procedure. An osteosynthesis was performed in 20 osteotomies (13 with screws and 7 with Kischner wires). The mean angle of osteotomy with a horizontal line was 13.5 ± 6.9 degrees. Average percentage of acetabular medial displacement was 63.7% ± 15.7%.

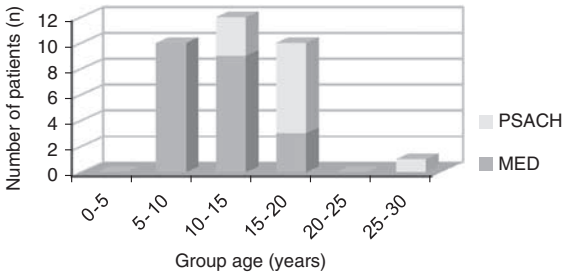

FIGURE 2. Distribution of the age at the time of the intervention in PSACH and MED groups. MED indicates multiple epiphyseal dysplasia; PSACH, pseudoachondroplasia.

Table 2 lists the measurements of the preoperative radiographs and of the data at last follow-up. A statistically significant improvement of all parameters was observed. The Sharp angle, center-edge angle and femoral head coverage evolved to a normal value at last follow-up.[21]

Most of the operated hips showed a Stulberg grade III or IV (aspherical congruency) (Figs. 3, 4). There was no difference between MED and PSACH groups ($P > 0.05$). We found no correlation between the age at the time of the intervention and the Stulberg grade at hip maturity or the final outcomes. Eight hips of 22 MED hips (36.4%) showed signs of AVN of the femoral head before Chiari osteotomy. These changes were unilateral in 4 patients and bilateral in 2.

The average PMA functional score improved significantly ($P < 0.001$) from 12.7 ± 2.9 (range, 3 to 15) preoperatively to 15.2 ± 3.1 (range, 9 to 18) at last follow-up. We received the answers to the HOOS survey from 11 patients (16 hips). The response rate was 73.3%. The mean age of these patients was 30.3 years and their mean follow-up since the Chiari osteotomy was 17 years, which is representative of the population of the study. The median HOOS worsens significantly over time, from 76 in patients with <15 years of follow-up to 45.4 in patients with >15 years of follow-up (P=0.04). In total, 17 of the 20 patients (85%) were satisfied with the results of Chiari osteotomy and would undergo it

TABLE 2. Radiographic Parameters Before the Chiari Osteotomy and at Last Follow-up

Radiographic Parameters	Initial (Mean ± SD)	Latest Follow-up (Mean ± SD)	Difference	*P*
Sharp angle (deg.)	47.7 ± 4.1	37.1 ± 4.4	−10.5	**<0.001**
Tönnis angle (deg.)	23.9 ± 7	16.9 ± 6.5	−7.8	**<0.001**
Center-edge angle (deg.)	12.3 ± 9.6	45.9 ± 11.5	−33.6	**<0.001**
Acetabular depth index	19.8 ± 4.5	22.4 ± 5.8	−2.5	**0.02**
Femoral head coverage (%)	62.6 ± 11.5	97.8 ± 15.5	+35.2	**<0.001**

Bold values indicate significant $P < 0.05$.

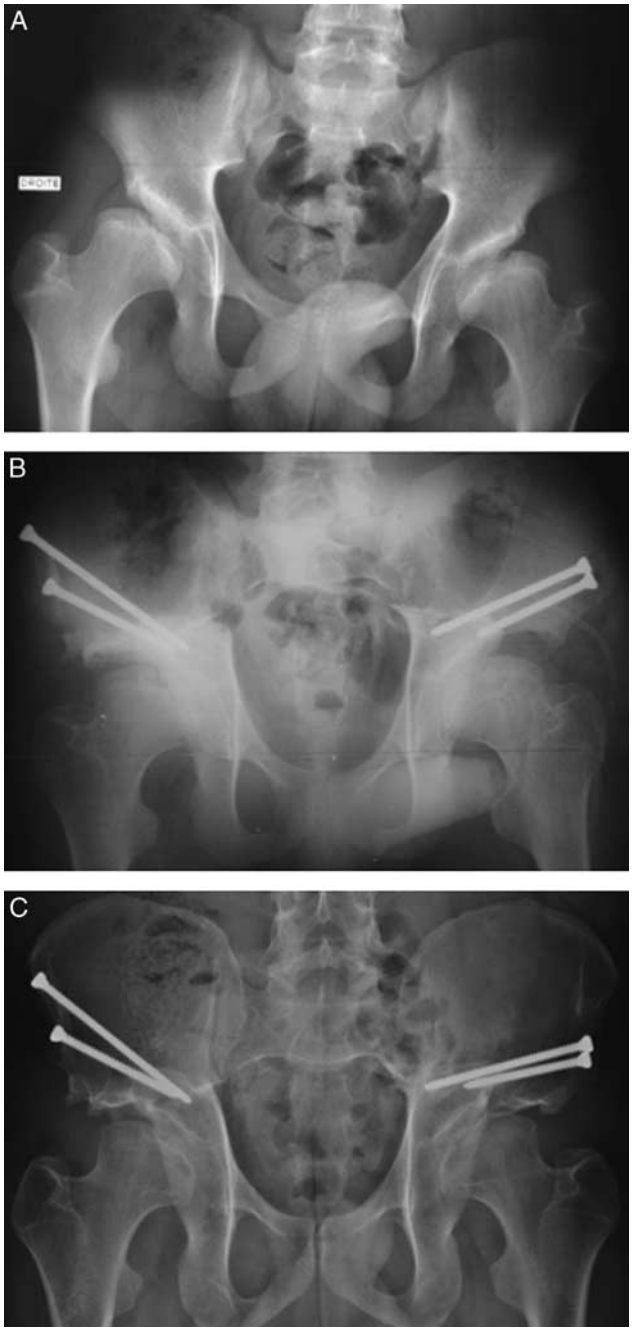

FIGURE 3. Radiographs of a multiple epiphyseal dysplasia male patient. A, Before Chiari osteotomy, at 16 years of age. B, Four months after right Chiari osteotomy and 1 day after left Chiari osteotomy. C, Seventeen years after bilateral Chiari osteotomy.

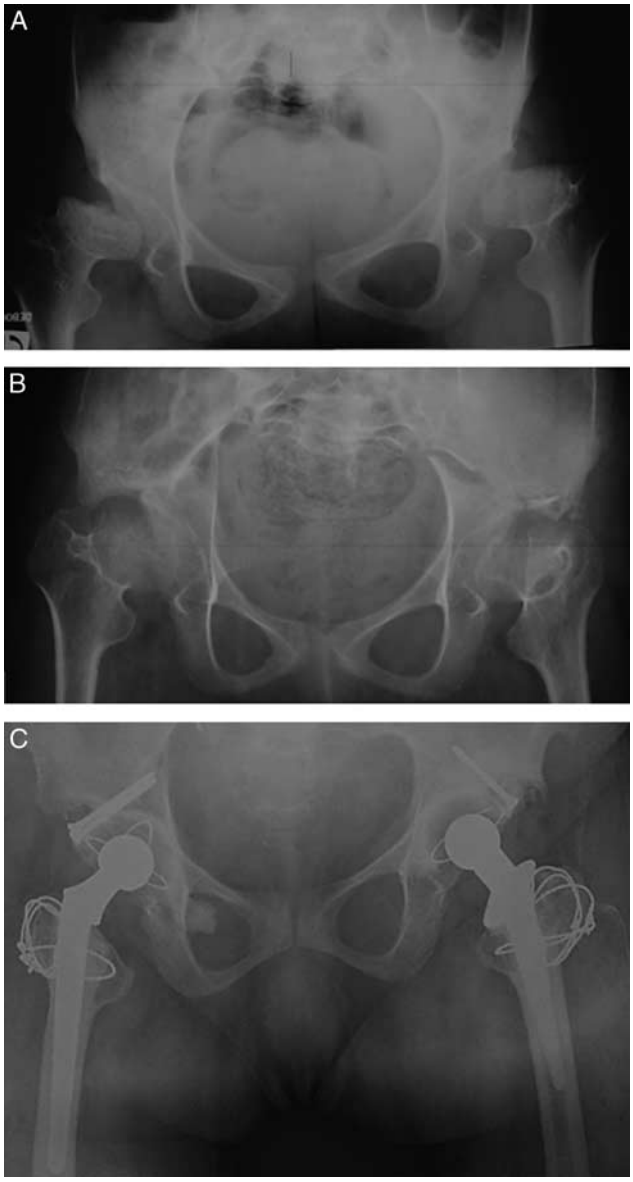

FIGURE 4. Radiographs of a pseudoachondroplasia girl. A, Before Chiari osteotomy. B, One year after bilateral Chiari osteotomy. C, She underwent total hip arthroplasty on the left side 35 years post-Chiari and on the right side 38 years post-Chiari.

again. For native hips (n=23), the outcomes according to Hosny and Fabry[17] at last follow-up were graded excellent in 10 hips (43.5%), good in 3 hips (13%), fair in 4 hips (17.4%), and poor in 6 hips (26.1%).

Seven minor complications, which did not require additional treatment, were noticed: lateral cutaneous nerve dysesthesia (n=3), heterotopic ossification (n=3), and secondary displacement (n=1). Conservative surgical revisions were performed in 3 hips of 2 MED patients: a bilateral triple pelvic osteotomy with a varus derotation femoral osteotomy and a core decompression and grafting for a femoral head AVN.

We determined the Treble index[5] for 20 MED hips, for which we had radiographs at the age of 10. Sixteen of these hips had a Chiari osteotomy and were graded type I —Sharp angle > 50 degrees, coverage of the femoral head inferior to 100%, epiphyseal index inferior to 0.3, and epiphyseal fragmentation. The evolution of these hips at skeletal maturity was: 1 congruent, 13 aspherical congruent, and 2 incongruent.

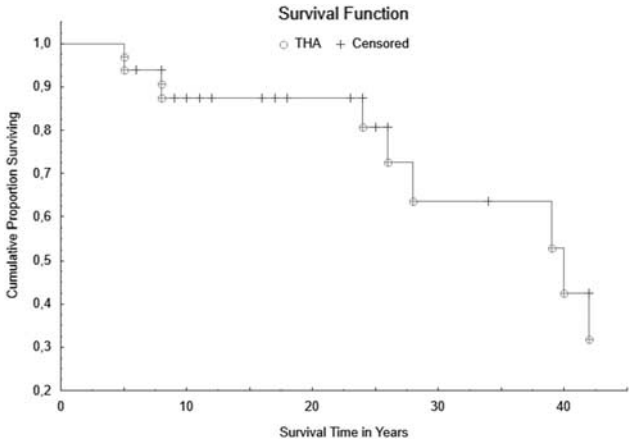

FIGURE 5. Survival analysis with total hip arthroplasty (THA) conversion as an endpoint.

At last follow-up, the evolution of 10 hips (30.3%) required THA in 7 patients (5 MED and 2 PSACH). Using the Stulberg classification,[20] 4 of them were graded III, 3 were graded IV, and 5 were graded V before hip arthroplasty. The mean age at the time of THA was 33.3 years (range, 18.2 to 53.3 y) at a mean post-Chiari interval of 19.5 years (range, 5.4 to 37 y). Survival analysis with THA conversion as the endpoint was 80.7% at 24 years postoperatively (95% confidence interval, 72.3%-89.1%) (Fig. 5). The survival rate was only 60% at 10 years for the 8 hips that presented signs of AVN in addition to the epiphysis anomalies.

DISCUSSION

The purpose of this article was to evaluate the results of Chiari osteotomy in MED and PSACH patients. To our knowledge, this is the first study to report long-term results of hip preservation surgery in these pathologies. The Chiari osteotomy is a procedure in which the capsule of the hip joint is used as an interposed soft tissue buffer between the femoral head and the shelf formed by division of the ilium.[12–14] The best indications for the Chiari osteotomy are painful dysplasia or uncoverage of the femoral head associated with incongruity or poor acetabulum development.[22,23] The advantages of this procedure are a load reduction on the femoral head by shortening the medial arm of the hip abductor lever system and immediate formation of a roof that is not absorbed. Only a few studies report the results of the Chiari osteotomy in children.[17,23,24] Unfortunately, none of these studies report long-term results. Hosny and Fabry[17] reported disappointing results in children under 10; this might be related to the anti-Chiari effect, which could be due to a damaged acetabular growth center. In a study about Perthes disease, Cahuzac et al[24] considered that Chiari osteotomy should only be done in children older than 8 years with incongruent, irregular, and subluxated hips. In our study, the anti-Chiari effect does not apply to MED or PSACH patients since there is usually no indication to operate on them before adolescence.

In our study, the osteotomy angle was slightly higher than 10 degrees and the mean percentage of displacement was 63.7%. This amount of displacement was required because of the high degree of dysplasia before surgery, with a mean femoral head uncoverage of 62.6%. The ideal Chiari osteotomy was described as marking an angle of 10 degrees upwards, immediately above the hip joint capsule and displaced by 50% of the pelvic width.[12,14] However, we did not observe a delay in consolidation in cases where the displacement was superior than 50% and there was no case of sciatic palsy, even transitory.

Our results showed that patient satisfaction was high; pain relief and improvement of function were constant. Most of radiographic parameters improved to normal values after the procedure. However, the majority of the operated hips showed a Stulberg grade III or IV at hip maturity. These good results degraded over time and 30.3% of the hips required THA at some point. The survival before conversion to THA was 80.7% at 24 years post-Chiari osteotomy, which is slightly inferior to the results reported in common hip dysplasia.[22,25,26]

The spontaneous evolution of the hip deformity in MED and PSACH children is known to lead to precocious osteoarthritis.[5–7] Treble et al[5] found that 9 of 10 MED patients with a type I hip showed clinical and radiologic signs of osteoarthritis before the age of 30. Moreover, AVN in a child with MED may result in additional structural deformity.[16] In our study, all the patients with AVN evolved to Stulberg grade III to V and half of them had a hip prosthesis before the age of 35. AVN of the femoral head in MED should be considered as a worse prognosis factor. Studies about the natural evolution of the hips in MED patients usually do not take into account the clinical and genetical heterogeneity of the pathology.[6] Seo et al[9] compared orthopaedic manifestations in MED patients and significantly found that abnormal development of the hip was commonly observed in the *COMP* group. They also found that the Stulberg grade was worse in that group than in the MATN3 group.

Sponer et al[11] reported satisfactory results after a Steel triple pelvic osteotomy in 3 patients and a short follow-up of 2 years. Li et al[10] reported good outcomes with 3 different surgical procedures (femoral intertrochanteric extension osteotomy, Staheli acetabular augmentation, and trochanter arthroplasty associated with Dega osteotomy) in a short series of 10 hips with a mean follow-up of 7.2 years. They recommended a trochanteroplasty to prevent a femoroacetabular conflict in severe hip deformities. We also use the triple pelvic osteotomy as a surgical option in MED patients younger than 10 years old with type I hips according to Treble or with AVN. We only have a follow-up of 10 years in 7 patients at present, but the results are encouraging since Stulberg grades of II and III exclusively are obtained. We do not propose the triple pelvic osteotomy in PSACH patients because of the severity of the deformations. Moreover, we do not think that another pelvic osteotomy would provide a good coverage of the femoral head in these patients.

A few studies reported the results of THA in skeletal dysplasia.[27–30] In this indication, THA is a technically challenging procedure with a high complication rate due the abnormal features of the hip, especially on the femoral side and because of the small height of these patients. Four studies

focused on the results of THA in MED. Ramaswamy et al[27] reported results with the longer follow-up (15.9 y) showing a high rate of revision (62.5%) and a poor postoperative PMA score reaching only 10.6 at last follow-up. No study currently exists for the PSACH patients. The Chiari osteotomy does not compromise a future THA[31] and is a better solution for symptomatic young patients with MED or PSACH.

This study has some limitations. It is a retrospective study about heterogeneous forms of MED and PSACH. The HOOS was not available preoperatively but PMA score and radiographies preoperatively and postoperatively were available for all the patients included. Even though we have the largest study of surgical treatment in MED and PSACH patients, we only reported on 33 operated cases, which limits the statistical significance of some results. Moreover, we did not have a control group. It should ideally be the next step to establish high level evidence guidelines of treatment but the low prevalence of these diseases is an issue for a case-control study.

The Chiari osteotomy provides good results in hip lesions caused by MED and PSACH. This intervention relieves pain, provides better function, and prevents early hip degradation. The very good long-term survival before conversion to THA should urge orthopaedists to first consider a conservative surgical procedure for these patients.

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