ORIGINAL PAPER

Triple osteotomy of the pelvis for Legg-Calve-Perthes disease: a mean fifteen year follow-up

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Abstract

Purpose This study presents the results of a prospective consecutive cohort of patients with Legg-Calvé-Perthes disease (LCPD) operated with triple osteotomy of the pelvis (TOP) between 1989 and 2005. We attempted to determine whether the results of TOP remain stable with time and consequently lower the risk of subsequent osteoarthritis. The primary study aims were to determine the maintenance of head coverage and joint congruity, and functional outcomes of this surgery.
Methods Forty-five patients with a mean follow-up of 15.2 years (range eight to 24) were included.
Results At latest follow-up, two patients were lost to follow-up, and two required a surgical reoperation. Cumulative maintenance of head coverage and joint congruity rate for all TOP was 84.6 % (95 % CI: 82.3–90.6 %) at 15 years. Factors significantly associated with poor long-term results were the age at diagnosis and Greene index.
Conclusion TOP in LCPD provides satisfactory and reproducible long-term clinical results.

Keywords Legg Calve Perthes · Osteotomy · Hip · Pediatrics

Introduction

The aim of the treatment of Legg-Calve-Perthes disease (LCPD) is to prevent hip osteoarthritis [1]. Many treatments have been described in order to prevent or limit femoral head deformation by containing the head within the acetabulum, using it as a mould for guiding its development. Some of the various procedures described (varisation osteotomy of the femur, Salter osteotomy) have sometimes resulted in a prolonged limp or inadequate containment [2]. Triple osteotomy of the pelvis (TOP) is now well documented [3–6]. Most of the available studies report very good results although with only short or medium-term follow-up.

We therefore addressed the following question: Do the results of TOP remain stable with time and consequently lower the risk of subsequent osteoarthritis?

The primary aims of this study were to determine the long-term radiographic results and functional outcomes and their determining factors

Methods

This study presents the results of a prospectively and consecutively accrued series of LCPD patients treated with TOP between 1989 and 2005. The indications for triple pelvic osteotomies included at least two of the following criteria: (1) six to ten years of age at clinical onset [7], (2) Catterall [8] groups III and IV, and Herring [9] lateral pillar groups B, B/C border, and C (Herring [9] type was considered after 1992), (3) loss of femoral head containment (subluxation) on the anteroposterior radiograph, (4) Catterall [8] signs of «head at risk». Exclusion criteria were: (1) joint stiffness, especially in abduction, (2) hinge abduction as systematically assessed by a hip arthrography under general anaesthesia.

All patients were operated upon by two surgeons (JSDG and JPC). The surgical technique has been previously described in literature [10, 11]. TOP combines a Salter innominate osteotomy and osteotomies of the iliopubic and ischiopubic rami. The surgery is performed under general anaesthesia on

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a standard surgical table. The child is installed in the three-quarter supine position, and the position is maintained by a sheet rolled against the child's back. The intervention begins with iliopubic and ischiopubic osteotomies via a genitofemoral approach. The innominate osteotomy follows, which is identical to a Salter osteotomy. The incision is a bikini-type incision located 1.5-cm below the iliac crest. It extends from the middle part of the iliac crest to the middle of the groin crease. The intermuscular space between the sartorius and tensor fascia lata is approached next. The osteotomy line is perpendicular to the wing of the ilium. It starts at the ischiatic notch and ends just above the antero-inferior iliac spine. A bicortical graft is harvested on the anterior and superior part of the wing of the ilium. The upper fragment is maintained, while the lower fragment is mobilized downward, outward, and forward. Displacement should be assisted with the Salter manoeuver, consisting of placing the heel on the knee of the contralateral knee. The graft is placed at the anterior opening and then fixed using two K-wires (diameter 15—18 depending on the child's age).

Baseline and perioperative information

Baseline information was collected from consenting patients and parents at the time of their pre-operative visit. Collected data included age, sex, age at diagnosis, delay of the diagnosis, duration of symptoms, Herring Score, Catterall score and Green Index [12]. This is the percentage of the width of the femoral head that is lateral to Perkins' line as determined from an AP radiograph of the pelvis. The amount of the ossific nucleus that is lateral to Perkins' line is measured in millimetres along a line perpendicular to Perkins' line. This distance is divided by the width of the opposite/normal femoral head as measured in millimetres along the epiphyseal plate. The quotient so obtained is multiplied by 100 to obtain the percentage of the femoral head that has been extruded from the acetabulum. Data were stored in a central database with the patients' and parents' consent.

Follow-up

Follow-up consisted of a review at baseline, three months, one year and two years postoperatively. Thereafter patients were followed yearly for ten years. This study reports latest follow-up outcomes.

Outcome measures

Patients were evaluated by an independent observer (RP). Complications and radiographic findings were recorded. The end point of the study was the latest follow-up or re-operation for any cause related to the hip. Clinical results were assessed by Oxford hip score (OHS) [13]. Plain anteroposterior pelvic

radiographs and a false lateral view according to Lequesne and de Sèze [14] were analysed. Lateral and anterior coverage were measured using the angles described by Wiberg [15] (lateral centre-edge angle) and Lequesne and de Sèze [14] (anterior centre-edge angle). The inclination of the acetabular roof was measured using the technique described by Tönnis [16] (acetabular index). The neck shaft angle (NSA) and the lateral extrusion index of Green [6, 12] were also assessed. The deformity index at two years described by Nelson et al. [17] was assessed. The severity of osteoarthritis at the last follow-up was graded from 0 to 4 according to Kellgren and Lawrence classification [18]. Heterotopic ossification was graded from 0 to 4 according to Brooker et al. [19]. Femoral head deformity and hip congruity were assessed by Stulberg classification [20].

Statistical analysis

Statistical analysis was carried out using EXCEL® (Microsoft Inc., Redmond, Wash.) and SPSS software® (SPSS Inc., Chicago, Ill). Descriptive data analysis was performed using Student t test. The difference between the preoperative and follow-up data was analysed with paired Student t tests. A Cox-proportional hazards model was used to compare the differences in TOP survival distributions for each of the co-variates recorded (patient age, gender, radiological parameters). A multivariate model was constructed and then covariates that were not significantly influential were systematically removed from the model to identify those having the greatest influence on survival. Significance was determined to be $p < 0.05$. Three Kaplan and Meier survivorship [21] analyses were performed using two different end points: an Oxford hip score less than 40 and Stulberg grade IV and V and/or Kellgren and Lawrence grade greater than 2.

Results

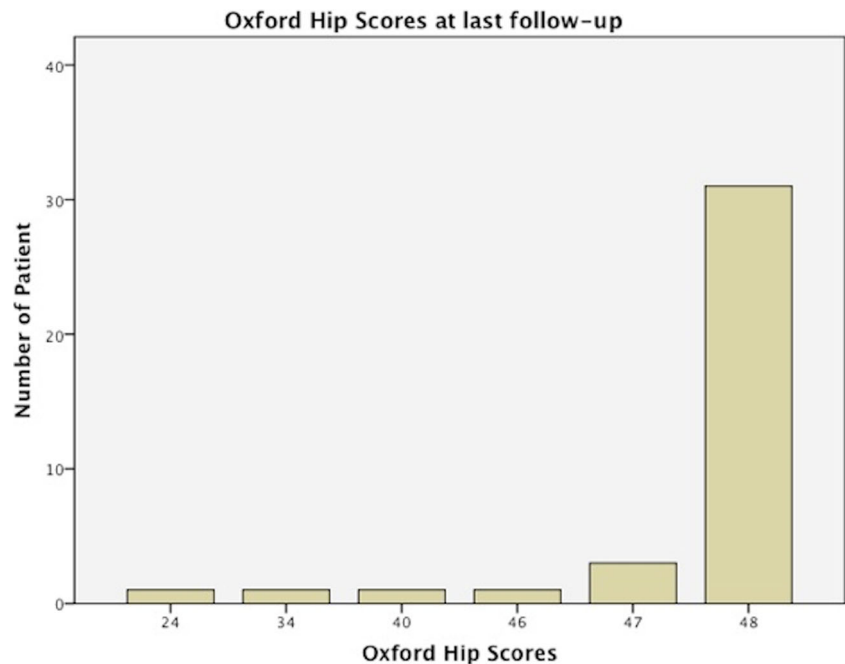## Descriptive analysis and factors affecting survival

After a mean follow-up of 15.2 years (range, 8.0–24.0) 45 patients were included with two patients lost to follow-up. There were 32 males and 11 females. The mean age at time of diagnosis was 6.1 years (range, three to 12) and delay of diagnosis was 3.4 months (range, 1.0–12.0). The mean duration of symptoms before surgical treatment was 6.6 months (range, 1.0–24.0). Mean age at surgery was 7.3 years (range, four to 12.4). The disease was bilateral in one case. Four patients (9.3 %) were rated Catterall 2, 25 (58.1 %) were Catterall 3 and 14 (32.6 %) were Catterall 4 (Table 1). Three patients (6.9 %) were rated Herring A, 26 (60.5 %) Herring B and 14 (32.6 %) Herring C.

Table 1 Descriptive analysis of potential risk factors

Characteristic	Minimum	Maximum	Mean	Standard deviation
Age at diagnosis (years)	2.5	12.0	6.14	2.12
Catterall	2.0	4.0	3.05	0.79
Green index	0	50.0	12.97	10.69
Deformity index	0.05	0.66	0.21	0.09
Duration of symptoms (months)	1.0	12.0	3.42	3.45
Delay of diagnosis (months)	1.0	24.0	6.65	4.38

Two patients, after a mean seven years from the index procedure, underwent a revision (JPC, JSDG). The first one was a male patient with a 2.5-cm symptomatic leg length discrepancy. The patient was treated with contralateral distal femoral physiodesis. The second was a male patient with a symptomatic retroverted femoral neck and coxa valga of 150°. The patient initially underwent a femoral osteotomy, then four years later a total hip arthroplasty (THA).

A Cox-proportional hazard model demonstrated a significantly increased risk of re-operation with a high Green index (risk of revision 0.147 for each increased percentage of Green index; $p=0.043$). Male gender was associated with a 0.048 increased risk of revision compared to female gender, although this was not statistically significant ($p=0.127$). Age of onset was associated with a higher risk of re-operation (0.417), but was not statistically significant ($p=0.183$).

Functional outcomes

The mean OHS at latest follow-up was 46.5 ± 4.5 (range, 24–48) (Fig. 1). There was no covariate influence on OHS except the age of onset which was associated with a 2.28 increased risk of OHS score <40 for each increased year of age ($p=0.035$). Only two patients (excluding the patient who underwent a total hip replacement) presented bad functional results with OHS <40. Thus survival rate for all TOP was 95.3 % (95 % CI, 90.3–98.6 %) at 15 years (Fig. 2) considering OHS <40 as an end point.

Radiological analysis

Coxometry parameters are presented in Table 2 at preoperative stage, postoperative stage, end of growth and at final follow-up (Figs. 3, 4, 5 and 6). There were neither statistical differences between postoperative and end of growth stage nor between end of growth stage and final follow-up. Only two patients presented osteoarthritis: one patient who required a THA was Kellgren and Lawrence 4 and the second was asymptomatic and rated Kellgren and Lawrence 3. Six patients (14 %) were Stulberg I, 19 (44 %) Stulberg II, 11 (25.5 %) Stulberg III, six (14 %) Stulberg IV and one (2.5 %) Stulberg V. Oxford hip score at latest follow-up was correlated with Stulberg classification with $r=0.446$ and $p=0.04$. Deformity index >0.3 was correlated with Stulberg grade III/IV with $r=0.786$ and $p=0.01$. There was no correlation between the preoperative Catterall score and the Stulberg score at the latest follow-up. Good radiological outcomes rate for all TOP was 84.6 % (95 % CI 82.3–90.6 %) at 15 years

Fig. 1 Distribution of Oxford hip score
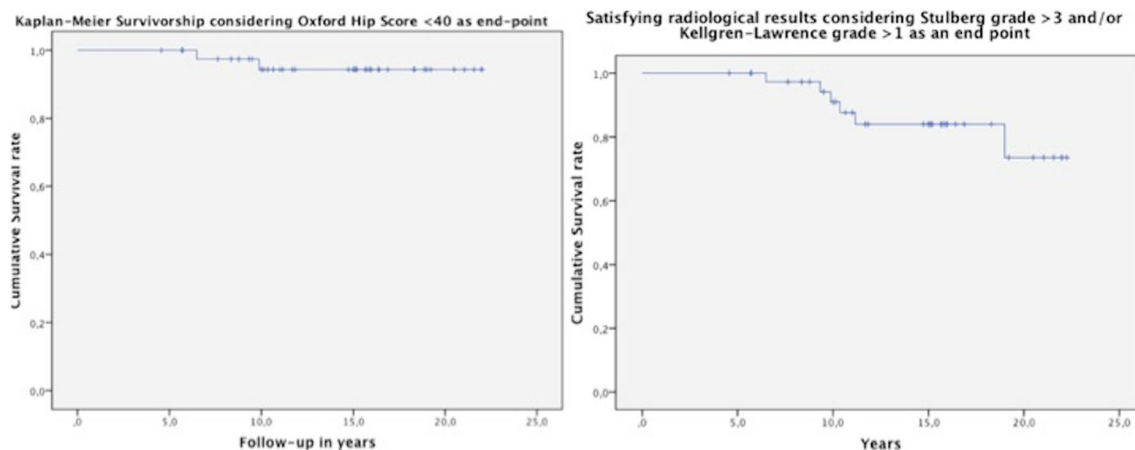
Fig. 2 Kaplan-Meier survivorship considering different endpoints

(Fig. 1) considering Stulberg IV and V and/or Kellgren Lawrence grade superior to 2, as an end point.

Discussion

Few studies to date have reported on the outcomes of triple osteotomy of the pelvis in LCPD with long-term follow-up. To the best of our knowledge, the present series represents the longest follow-up cohort described in literature. This study demonstrates very good long-term survival after TOP with a rate of good radiological outcomes (Stulberg 1 to 3 and/or Kellgren Lawrence<2) of 84.6 % (95 % CI 82.3–90.6 %) at 15 years.

The present study has some limitations. First, two patients (4.4 %) were lost to follow-up. Second, the Cox proportional hazard modelling did not take into account the learning curve of the surgeon, which may have skewed failure rates. Overall, the power of the study is limited by the small number of patients (43 patients) and may not allow a significant risk factors analysis. Finally, this study because of the limited number of patients presents a risk of type 2 error, and many prognostic factors considered important in other Perthes' studies [7] may have been overlooked.

In the present series, absence of hinge abduction was systematically assessed by hip arthrography under general anaesthesia before any procedure since it was considered as a contra-indication to TOP. Indeed, established hinge abduction due to enlarged, crushed femoral head may lead to an irreducible hip, and further attempts to contain the femoral head may be more harmful than helpful [22–25]. Furthermore, in this study all the procedures were performed at fragmentation stage. Herring et al. [26] demonstrated that femoral head was all the more likely to undergo progressive flattening in older patients, with severe lateral pillar involvement, and in those with delayed reossification. Joseph et al. [27] concluded that containment surgery aiming at preventing femoral head deformation in LCPD should be performed before the advanced stage of fragmentation.

Thus, time to surgery is a prognostic factor of an utmost importance for TOP outcomes. Considering risk factors associated with functional outcomes, only Green Index and age of the patient at time of diagnosis seemed to be significant in our study. Green et al. [12] initially developed the femoral epiphyseal extrusion index as a prognostic indicator in LCPD. Authors noted 20 % and less protrusion as a good prognostic indicator in LCPD cases. Tannast et al. [28] alluded to an extrusion index of less than 25 % as normal, although they emphasized no study has defined a minimum extrusion. Hosalkar et al. [6] also observed that extrusion index of less than 20 % was a good prognostic indicator. As Nelson et al. [17], we found that deformity index>0.3 at two years postoperative was also a good prognosis factor of bad radiological

Table 2 Descriptive analysis of radiographic parameters

Measure	Preoperative		Postoperative		End of growth		Last follow up	
	Mean	Standard deviation	Mean	Standard deviation	Mean	Standard deviation	Mean	Standard deviation
Lateral centre-edge angle	41.0	9.05	46.9	12.05	45.0	9.17	37.5	6.66
Acetabular index	10.6	4.42	8.9	4.59	9.4	4.38	13.6	4.55
Anterior centre-edge angle	39.1	5.54	40.5	7.15	36.5	3.17	37.5	3.15
NSA	135.6	4.44	137.6	5.13	137.7	5.10	136.9	5.43

Fig. 3 Box-plots of pre-operative, postoperative and last follow-up acetabular index
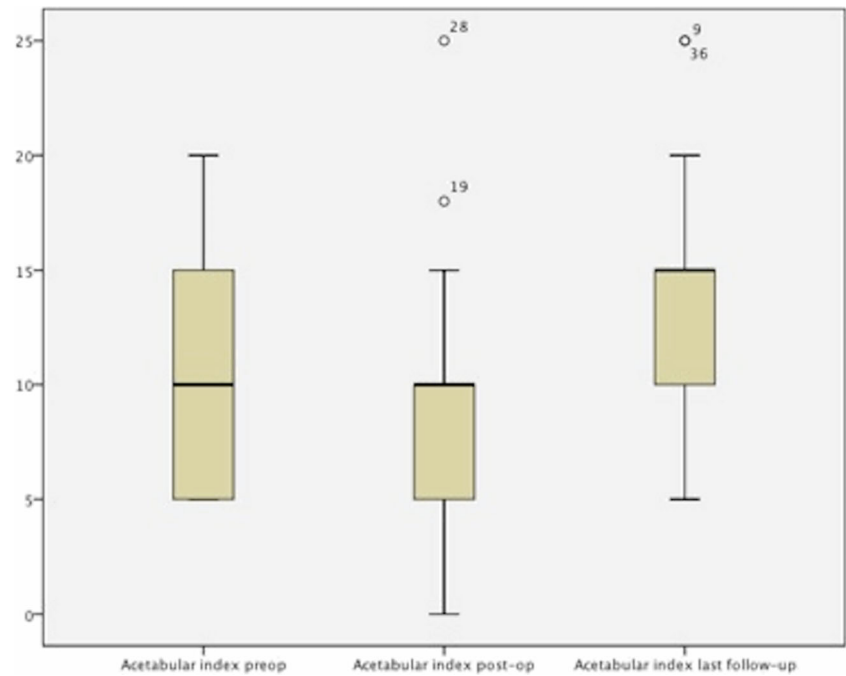

outcome. More recently, Kwang-Won Park et al. pointed out that premature physeal closure of the proximal femoral physis in patients with LCPD occurred 3.5 years earlier than normal hips and could be an adjunct diagnostic tool in the prognostication of LCPD outcomes [29].

Although age of onset was not significant here, Wenger et al. reported in a series of 39 patients, that 17 % of lateral pillar C patients below the age of eight had a poor outcome compared with 50 % above the age of eight [30].

Our study is consistent with the good results of series with short to medium-term follow-up using the same surgical technique. Wenger et al. reported 89 % good results after TOP in a series of 21 patients [30]. Conroy et al. performed TOP in eight patients with a minimum follow-up of four years [4]. The modified Harris hip score (HHS) ranged from 38 to 60 pre-operatively and from 77 to 92 postoperatively. O'Hara et al. reviewed 21 patients after TOP with a 30-month follow-up (one to five years) and found very good results [31].

Fig. 4 Box-plots of pre-operative, postoperative and last follow-up lateral centre-edge angle
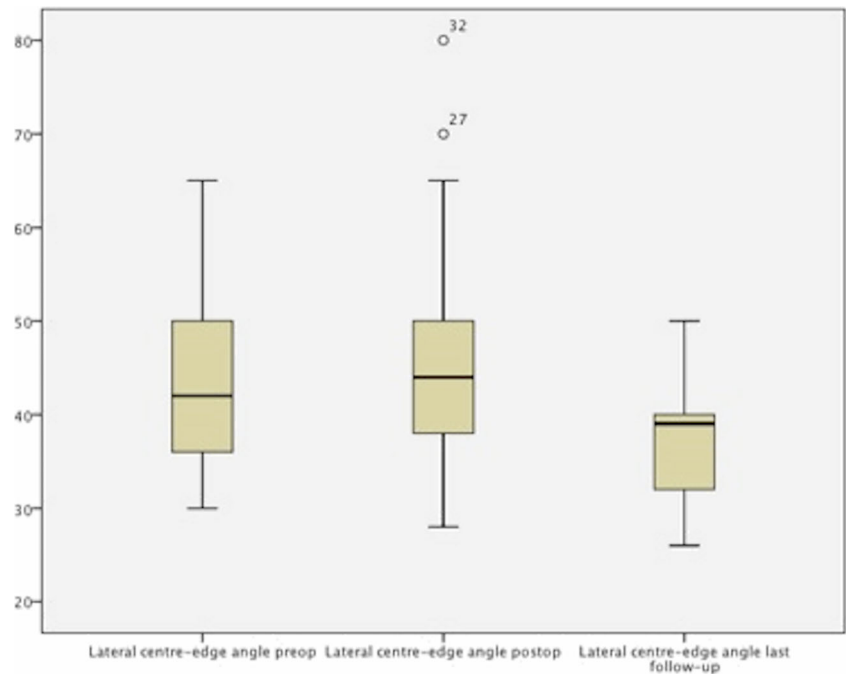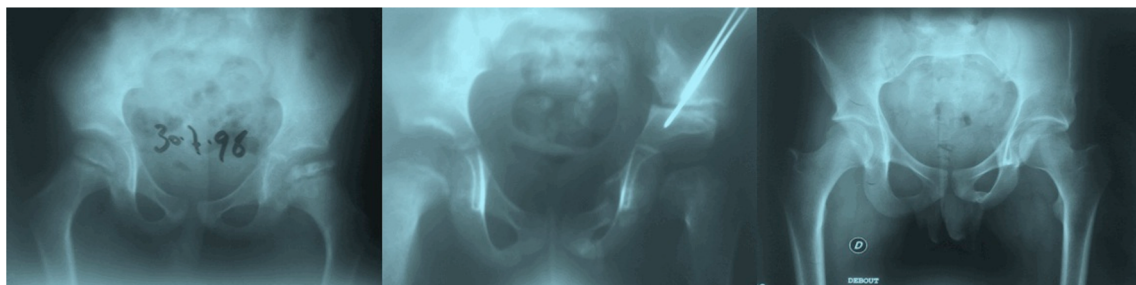
Fig. 5 Box-plots of pre-operative, postoperative and last follow-up anterior centre-edge angle
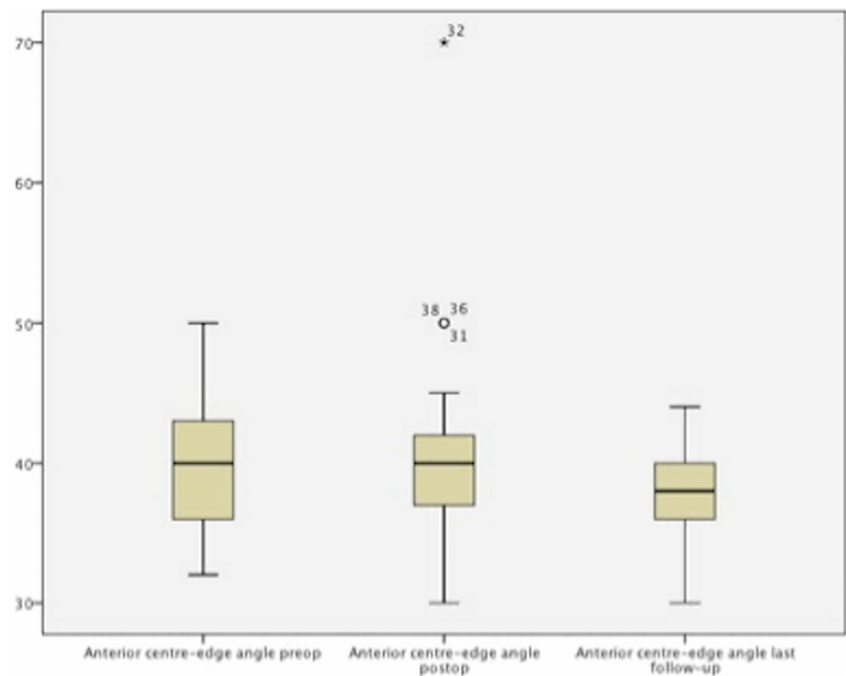
The average gain in HHS was 34.3 (range four to 55). Vukasinovic et al. also reported very good results in a series of 30 patients with mid-term follow-up [32]. More recently, Hosalkar et al. demonstrated the durability of femoral head containment with TOP in a series of 20 patients with a minimum follow-up of three years [6].

Nonoperative treatment remains an alternative for Catterall 1 et 2. Recently, Larson et al. [33] reported on the 20-year follow-up results of a prospective multcentric study of nonoperative treatment and found a high prevalence of osteoarthritis and low clinical outcome scores. Approximately 20 % of Stulberg type II hips and 60 % of type III, IV, and V hips showed radiographic evidence of osteoarthritis. As in our series, a high Stulberg grade was associated with pain, impingement on physical examination and arthritis. Similarly, McAndrew and Weinstein displayed a 50-year follow-up of the Gower and Johnston historical series [34]. By that time, 15 (39 %) of the 35 patients had required THA, and six (21 %) had an IHS (Iowa hip score) <80.

Other authors have recommended varus femoral osteotomy. Terjesen et al. reported in a prospective study on 70 patients that for children aged six to ten years, in whom the whole femoral head was affected, femoral head sphericity five years after femoral osteotomy was better than for those having undergone physiotherapy [35]. However, Mirovsky et al. [36] reported a 58 % incidence of leg length discrepancy (LLD). Grzegorzewski et al. [37] stated that varus osteotomy produces more shortening than other methods of treatment. Van Der Geest et al. advocated shelf acetabuloplasty for severe cases of LCPD, based upon a retrospective series of 30 patients with 12 years follow-up [38]. Bernese's (Ganz) periacetabular osteotomy with lateral muscle sparing has more recently emerged and gained popularity around the globe [2, 39]. However, to date, there is no high level evidence study to assess outcomes. Salter osteotomy in LCPD is an effective treatment which may alter the natural course of the disease. Similar to TOP, its main advantage is its effect on femoral head remodelling with remaining growth [7, 40]. Nonetheless,

Fig. 6 Pre-operative, postoperative and 16-years follow-up radiographs of the same patients

this osteotomy has some limitations in patients above six years old given that appropriate tilt may not be achieved due to the relative stiffness of the pubic symphysis.

In addition to the deformities caused by the disease process, containment procedures may cause femoro acetabular impingement (FAI). For instance, Salter osteotomy, shelf procedure, or TOP may create pincer-type impingement [41, 42] which is a pre-arthritic condition [28, 43]. Therefore risk-benefit ratio must be taken into account before considering any surgical containment. Furthermore, it is unclear whether and to what extent the acetabular coverage changes during growth and after TOP. Hosalkar et al. [6] recommended an acetabular coverage after TOP for LCPD (in Catterall Stage III or IV) of 44° or less and an ARA below –6°, as they noted residual remodeling could allow a return to a nonpincer morphology with growth. Therefore, in growing patients presenting asymptomatic pincer morphology, it seems reasonable to carry on with follow-up until completion of skeletal maturity [6, 44]. According to Hosalkar et al. [6], aggressive procedure aiming at deimpingement may not be necessary unless the patient has severe symptoms. In our study, one patient presented clinical signs of impingement with a cross-over sign indicating a retroverted acetabulum, possibly related to the pelvic osteotomy.

Although TOP may delay secondary osteoarthritis, the resulting altered anatomy of the pelvis and proximal femur might lead to a technically challenging THA [45]. Controversy remains on this point as some investigators have shown a possible worsening of the outcome of THA after a TOP, whereas others found it did not compromise the results of THA if TOP is properly performed [39, 46, 47]. In a recent study, Tokunaga et al. [48] demonstrated that prior pelvic osteotomy lead neither to a higher peri-operative complication rate, nor a higher revision rate, nor compromised HHS nor shortened survivorship after THA.

TOP resulted in maintenance of head shape in lateral pillar B or C patients with a cumulative survival rate of 95.3 % (95 % CI 90.3–98.6 %) at 15 years in this series This method provides effective containment, which allows prolonged remodeling while avoiding the limitations of femoral varus osteotomy (limp, short limb) and Salter osteotomy (incomplete containment).

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