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Research article

# The acetabulum in healed Legg–Calvé–Perthes disease is cranially retroverted and associated with global reduction of femoral head coverage: a matched-cohort study

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## ABSTRACT

To evaluate the acetabular morphology in healed Legg–Calvé–Perthes disease after skeletal maturity using computed tomography (CT) scan and to compare with matched controls. We identified 33 (37 hips) patients with healed Legg–Calvé–Perthes disease and closed triradiate cartilage who underwent pelvic CT scan. Each patient was matched based on sex, age and side to a subject with no history of hip disease who had undergone pelvic CT evaluation because of abdominal pain. Both cohorts had 23 (70%) males and mean age of 16.4–16.5 $\pm$ 3.6 years. Two independent readers assessed lateral center-edge angle (LCEA), acetabular inclination angle (IA), acetabular depth-width ratio (ADR), acetabular version 10 mm below the dome (cranial) and at the acetabular center and anterior (AASA) and posterior acetabular sector angles (PASA). All measurements had good to excellent interobserver agreement (intraclass coefficients $\geq$ 0.87). The hips in the Legg–Calvé–Perthes disease cohort had a smaller mean $\pm$ standard deviation (SD) superior, anterior and posterior acetabular coverage as assessed by LCEA (13.2° $\pm$ 10.7° versus 28.2° $\pm$ 3.4°; $P < 0.0001$), IA (11.6° $\pm$ 6.7° versus 3.5° $\pm$ 2.8°; $P < 0.0001$), AASA (52.4° $\pm$ 9.5° versus 59.3° $\pm$ 5.0°; $P = 0.001$) and PASA (79.3° $\pm$ 5.9° versus 92.3° $\pm$ 5.5°; $P < 0.0001$) compared with controls. The acetabulum was shallower (ADR 287 $\pm$ 45 versus 323 $\pm$ 28; $P = 0.0002$) and the acetabular version was decreased cranially (0.4° $\pm$ 9.2° versus 8.2° $\pm$ 6.8°; $P = 0.0002$) and at the acetabular center (13.7° $\pm$ 5.1° versus 17.2° $\pm$ 3.8°; $P = 0.004$) in Legg–Calvé–Perthes disease hips. After skeletal maturity, hips with healed Legg–Calvé–Perthes disease have shallower and more cranially retroverted acetabula, with globally reduced coverage of the femoral head compared with age-, sex- and side-matched control hips.

## INTRODUCTION

Legg–Calvé–Perthes disease [1–3] typically heals with varying degrees of hip deformities [4]. Previous studies have focused on the description of the anatomical abnormalities of the femur [5, 6]. However, the acetabular morphology may also influence the long-term prognosis of the disease [5, 7, 8]. Acetabular deformities described in Legg–Calvé–Perthes disease include acetabular dysplasia [5, 7, 9, 10], acetabular retroversion [6, 11–15], compartmentalization of the articular surface [5, 16, 17], irregularity of the acetabular contour [5, 9] and flattening or sloping acetabular margin [18, 19]. These acetabular deformities combined with the typical aspherical femoral head, short femoral neck and high-riding greater trochanter may lead to hip pain associated with structural hip instability due to insufficient femoral head coverage [20] or femoroacetabular impingement [15, 21, 22]. Understanding the complex acetabular deformity in Legg–Calvé–Perthes disease is important for the evaluation and treatment of patients with symptoms of the healed hip. However, the patterns of acetabular deformity are not well understood.

The purpose of this study was to compare the acetabular morphology after the skeletal maturity of patients with healed Legg–Calvé–Perthes disease who had not undergone surgical treatment, with sex-, age- and side-matched subjects without a history of hip disease, using computed tomography (CT) imaging.

## MATERIALS AND METHODS

This retrospective matched-cohort study was approved by our ethics and institutional review board committee, with waiver of informed consent. From January 2000 to June 2017, 496 patients with Legg–Calvé–Perthes disease were seen in our Institution. Of those, 95 (19%) patients underwent a pelvic CT for 3D evaluation of the hip. Thirty-nine of them were excluded because of prior hip surgery including femoral or pelvic osteotomies (33 patients), concurrent genetic syndrome (two patients) or spastic diplegia (one patient), unilateral hip CT (two patients) and movement artifacts on the CT images (one patient). Twenty-three patients were excluded because of the presence of open triradiate cartilage on CT scans. The complete closure of the triradiate cartilage was necessary to warrant assessment of completely ossified acetabular walls for measurements of acetabular morphology [23, 24]. All hips were categorized as late stage according to the Waldenström classification [25], using radiographs obtained at the same time as the CT acquisition. The final cohort included 33 patients with healed Legg–Calvé–Perthes disease [25], four of them with bilateral disease, yielding a total of 37 hips. The control subjects were non-syndromic patients without hip-related symptoms, who had a pelvic CT for assessment of clinical presentation of acute abdominal pain and the suspicion of appendicitis, and closed triradiate cartilage on CT scans. The control cohort comprised individuals matched 1-to-1 with the patients in the Legg–Calvé–Perthes disease cohort by sex and age within a maximum difference of 9 months, using a nearest-neighbor approach. We finally matched the 37 control hips according to the affected side of Legg–Calvé–Perthes disease patients.

The Legg–Calvé–Perthes disease and the matched cohort each had 23 (70%) male and 10 (30%) female subjects (Table I). The mean age (and standard deviation) was 16.4 ± 3.6 years for Legg–Calvé–Perthes disease cohort (range 12.3–25.6 years) and 16.5 ± 3.6 years for the matched cohort (range 12.2–26.0 years). All hips were further classified according to Stulberg *et al.* [8] (Table I).

**Table I. Characteristics of Legg–Calvé–Perthes and matched control groups (33 patients; 37 hips per each group)**

| | *Perthes* | *Controls* |
|---|---|---|
| Age (years) | 16.4 ± 3.6 (12.3–25.6) | 16.5 ± 3.6 (12.2–26.0) |
| Sex (% male) | 23 (70) | 23 (70) |
| Involved side | | |
| Right | 18 (55) | 18 (55) |
| Left | 11 (33) | 11 (33) |
| Bilateral | 4 (12) | 4 (12) |
| Waldenström stage ($n = 37$ hips) | | |
| Late | 37 (100) | |
| Stulberg *et al.* classification ($n = 37$ hips) | | |
| I | 0 (0) | |
| II | 2 (5) | |
| III | 12 (32) | |
| IV | 22 (59) | |
| V | 1 (3) | |

Values are expressed in mean, standard deviation (SD) and range (age) or frequency and percentage (other variables).

### Acetabular morphology assessment on CT imaging

Pelvic CT images were obtained with patients in supine position and hips in extension, and neutral abduction, adduction and rotational positioning. Coronal and axial reformats through the centers of the femoral heads were created from thin-section axial datasets using 2D multiplanar reconstructions (OsiriX, Version 5.8.2, Pixmeo, Bernex, Switzerland). We identified specific symmetry of anatomic landmarks to correct for variations in pelvic tilt and rotation in the coronal and axial planes. The reformatted coronal plane was identified as the plane of the centers of both femoral heads and the apex of the acetabulum dome. One-mm axial reformats were acquired directly orthogonal to the reformatted coronal plane [26].

In the coronal plane passing through the center of the femoral heads, we measured the lateral center-edge angle (LCEA) as described by Wiberg [27] (*Figure 1A*), using the lateral sclerotic edge of the sourcil as the reference for

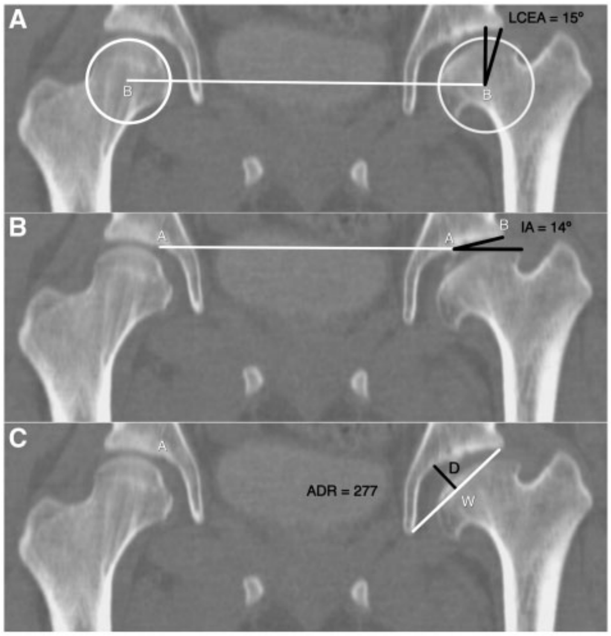


**Fig. 1.** Coronal CT reconstruction passing through the center of the femoral heads of the pelvis of a 14-year-old boy with healed Legg–Calvé–Perthes disease on the left hip. The right hip was not affected by the disease. (**A**) The lateral center-edge angle was assessed using a line perpendicular to the line (BB) connecting the center of the femoral heads (B) or acetabulum (Visser center) and a line from the most lateral point of the acetabular sourcil to the center of the hip (B). (**B**) The acetabular IA was assessed using a transverse line transecting the medial limit (Point A) of the acetabular sourcil of both hips, and a line connecting the medial (Point A) and lateral (Point B) aspects of the acetabular sourcil. (**C**) The acetabular depth-width ratio was calculated as depth/width × 1000. The acetabular width (W) was measured from the most lateral point of the sourcil to the most lateral inferior point of the teardrop. Acetabular depth (D) was measured from the midpoint of the connecting W line to the deepest point of the acetabular fossa.

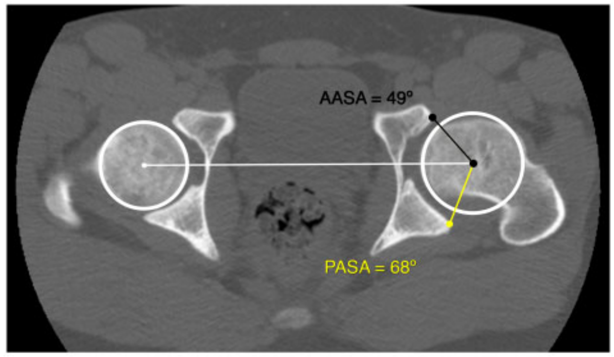


**Fig. 2.** Axial CT reconstruction passing through the center of the femoral heads of the pelvis of a 16-year-old boy with healed Legg–Calvé–Perthes disease on the left hip. The anterior acetabular sector angle was assessed using a transverse line connecting the centers of both femoral heads (white line) and a line connecting the anterior margin of the acetabulum to the center of the femoral head (black). The posterior acetabular sector angle was assessed using the transverse line (white) connecting the posterior margin of the acetabulum to the center of the femoral head (yellow). The right hip was not affected by the disease.

superior acetabular coverage [28]. The center of the femoral head for concentric hips or the center of the acetabulum was used as the point of reference for non-spherical femoral heads or non-concentric hips, as described by Visser [29]. The acetabular inclination angle (IA) was assessed using the acetabular roof angle as described by Tönnis [30] (*Figure 1B*). The acetabular depth-width ratio (ADR) [7] was assessed in the reformatted coronal plane as described by Fujii *et al.* [31] (*Figure 1C*).

In the axial plane, we assessed the anterior and posterior acetabular support by measuring the anterior (AASA) and posterior (PASA) acetabular sector angles [32] (*Figure 2*), wherein the Visser's center [29] was used for non-spherical femoral heads or non-concentric hips. The cranial acetabular version (*Figure 3A*) was measured at the level 10 mm caudal to the acetabular dome [26, 33], and the central acetabular version (*Figure 3B*) was measured using the CT slice passing through the femoral head centers [34] or the Visser's center [29]. The centers were determined on the coronal plane, using multiplanar orientation [26]. Two pediatric orthopedic surgeons (DAM with 9 years, and MF with 4 years of practice), who were not involved in the clinical care of the patients, measured all parameters for each hip included in this study, using an independent and blind approach.

### Statistical methods

Intraclass correlations (ICC) were assessed using a two-way analysis of variance model for absolute agreement with two fixed readers. Values of ICC (and 95% confidence intervals) were good to excellent [35] (Table II), and the measurements of the two readers were averaged for further analyses. Legg–Calvé–Perthes disease hips and control hips were compared with a two-sample paired *t*-test. As an effect of the matching, adjustment for sex, age and side had little effect in the analysis, and we report only the unadjusted results.

## RESULTS

All measurements of acetabular morphology differed between healed Legg–Calvé–Perthes disease and matched cohorts, indicating that in Legg–Calvé–Perthes hips there was a global reduction in acetabular coverage of the femoral head and acetabular retroversion (Table III and Figure 4).

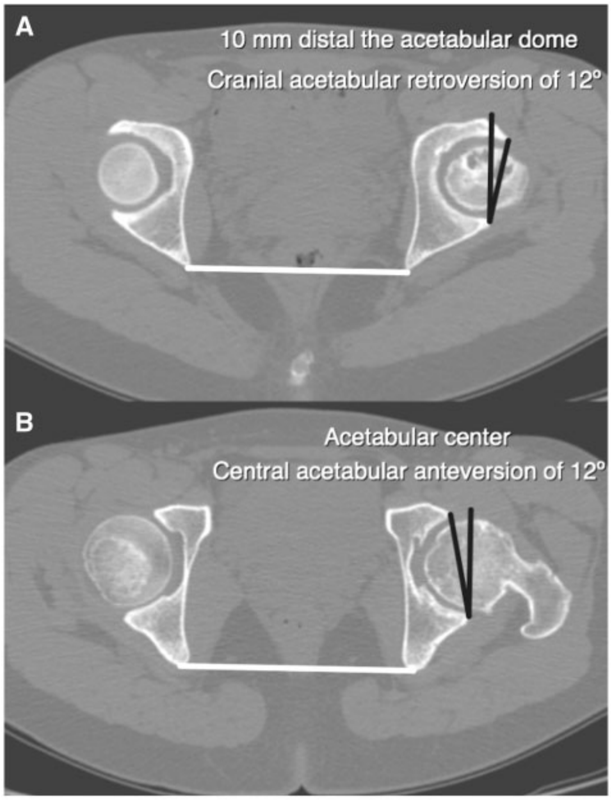


**Fig. 3.** Axial CT images of the pelvis of a 16-year-old girl with healed Legg–Calvé–Perthes disease on the left hip. The right hip was not affected by the disease. (**A**) Axial CT slice was acquired 10 mm distal to the acetabular dome using multiplanar guidance. The cranial acetabular version was the angle (black) formed by a line that is perpendicular to the horizontal line connecting the most posterior aspect of both acetabula (white line) and a line connecting the anterior and posterior margins of the acetabulum. (**B**) Axial CT slice was acquired at the level of the centers of the femoral heads using multiplanar guidance. The central acetabular version was the angle (black) formed by a line that is perpendicular to the horizontal line connecting the most posterior aspect of both acetabula (white line) and a line connecting the anterior and posterior margins of the acetabulum.

Decreased lateral coverage of the femoral head as assessed by smaller mean LCEA was found in Legg–Calvé–Perthes subjects compared to controls (mean ± SD, 13.2° ± 10.7° compared with 28.2° ± 3.4°, $P < 0.0001$). Combined with higher values of IA (mean ± SD, 11.6° ± 6.7° compared with 3.5° ± 2.8°, $P < 0.0001$), these findings suggested acetabular dysplasia. In the Legg–Calvé–Perthes disease group, reduced depth of the acetabulum with a global reduction of head coverage was demonstrated by reduced ADR (mean ± SD, 287 ± 45 compared with 323 ± 28, $P = 0.0002$), lower mean values of AASA (mean ± SD, 52.4° ± 9.5° compared with 59.3° ± 5.0°, $P = 0.001$) and PASA (mean ± SD, 79.3° ± 5.9° compared with 92.3° ± 5.5°, $P < 0.0001$) in comparison with hips from control subjects. The acetabular version was significantly lower in Legg–Calvé–Perthes disease hips both cranially (mean ± SD, 0.4° ± 9.2° compared with 8.2° ± 6.8°, $P = 0.0002$) and at the center of the femoral head (mean ± SD, 13.7° ± 5.1° compared with 17.2° ± 3.8°, $P = 0.004$), suggesting acetabular retroversion. Although the difference in cranial acetabular version between Legg–Calvé–Perthes disease and control cohorts was of 7.9°, this was larger than the 3.4° difference in the acetabular version at the center of the acetabulum (paired *t*-test on difference of differences, $P = 0.002$), indicating that retroversion was more pronounced at the cranial part of the acetabulum.

**Table II. Reliability of acetabular measurements**[a]

| *Acetabular measurement* | *ICC* | *95% CI* |
|---|---|---|
| Lateral center-edge angle | 0.91 | 0.86–0.95 |
| Acetabular inclination angle | 0.93 | 0.87–0.95 |
| Acetabular depth-width ratio | 0.88 | 0.81–0.92 |
| Anterior acetabular sector angle | 0.90 | 0.82–0.95 |
| Posterior acetabular sector angle | 0.92 | 0.86–0.96 |
| Cranial acetabular version | 0.87 | 0.80–0.92 |
| Central acetabular version | 0.91 | 0.85–0.94 |

[a]Measurements of hips in the Legg–Calvé–Perthes disease and matched control cohorts. The agreement between two observers was assessed using intra-class correlations (ICCs) with 95% confidence intervals (CIs). A two-way analysis of variance model for absolute agreement was used. The cranial acetabular version was measured 10 mm caudal to the acetabular dome. The central acetabular version was measured at the level of the center of the femoral heads or Visser's centers for non-concentric or non-spherical hips.

No differences in acetabular measurements were found between the Stulberg *et al.* II and III categories (sphericity and congruence partially preserved) in comparison with IV and V (coxa plana or incongruent hips) (Table IV).

## DISCUSSION

Legg–Calvé–Perthes disease may lead to deformity of the proximal femur and acetabulum, and despite the fact that femoral deformities have been well-described, the acetabular morphology is not completely understood [6]. After the healing stage of Legg–Calvé–Perthes disease, the progression to hip osteoarthritis depends on the articular morphology and congruency; therefore, it is essential to understand the complex anatomy of the acetabulum [5, 7, 8, 22]. In the present study, we found that the

**Table III. Comparison of acetabular morphology between the Legg–Calvé–Perthes and matched cohorts**

| | *Cohorts*[a] | | | |
|---|---|---|---|---|
| *Acetabular measurements* | *LCPD* | *Control* | *Difference (95% CI)* | P-*value* |
| Lateral center-edge angle (°) | 13.2 ± 10.7 | 28.2 ± 3.4 | −15.0 (−18.6 to −11.4) | <0.0001 |
| Acetabular inclination angle (°) | 11.6 ± 6.7 | 3.5 ± 2.8 | 8.1 (5.6–10.6) | <0.0001 |
| Acetabular depth-width ratio | 287 ± 45 | 323 ± 28 | −36 (−54 to −18) | 0.0002 |
| Anterior acetabular sector angle (°) | 52.4 ± 9.5 | 59.3 ± 5.0 | −6.9 (−10.8 to −3.0) | 0.001 |
| Posterior acetabular sector angle (°) | 79.3 ± 5.9 | 92.3 ± 5.5 | −12.9 (−15.7 to −10.1) | <0.0001 |
| Cranial acetabular version (°) | 0.4 ± 9.2 | 8.2 ± 6.8 | −7.9 (−11.7 to −4.0) | 0.0002 |
| Central acetabular version (°) | 13.7 ± 5.1 | 17.2 ± 3.8 | −3.4 (−5.7 to −1.1) | 0.004 |

[a]Acetabular measurements in hips with Legg–Calvé–Perthes disease (LCPD, $n = 37$ hips) and matched control ($n = 37$ hips) cohorts summarized by mean and standard deviation. The differences with 95% confidence interval (CI) between the cohorts are given, whereas positive difference indicates higher means in the LCPD cohort. Cranial acetabular version was measured at a level 10 mm caudal to the acetabular dome, and central acetabular version at the level of the center of the femoral heads or Visser's centers in non-concentric or non-spherical hips.

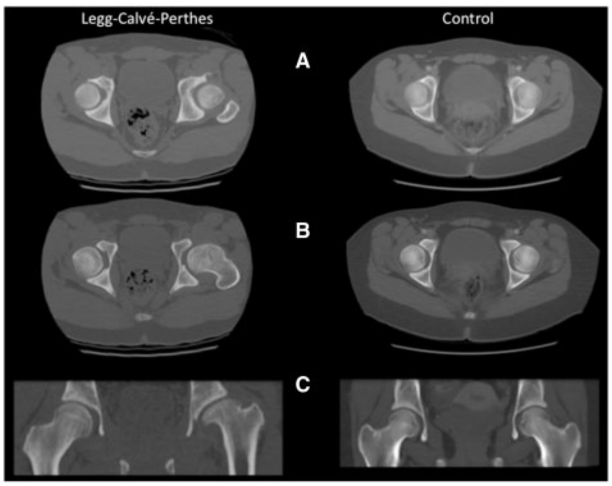


**Fig. 4.** CT reconstruction of the pelvis of a 16-year-old boy with a healed Legg–Calvé–Perthes disease on the left hip and a 16-year-old boy included in the control group. (**A**) Axial CT images of the pelvis 10 mm distal to the acetabular dome. The acetabulum in Perthes disease was more retroverted compared with control. (**B**) Axial CT reconstruction passing through the center of the femoral heads of the pelvis. The acetabulum in Perthes disease was anteverted but showed less anterior and posterior coverage of the femoral head compared with control. (**C**) Coronal CT reconstruction passing through the center of the femoral heads of the pelvis. The acetabulum in Perthes disease was dysplastic and shallower compared with control.

acetabulum in Legg–Calvé–Perthes disease is, on average, cranially retroverted, dysplastic and shallow in comparison with matched control hips from patients without hip conditions.

Acetabular dysplasia in Legg–Calvé–Perthes disease has previously been reported in the literature [5, 10, 20, 36]. Our findings indicate that the acetabulum is dysplastic and provides less anterior, superior and posterior coverage to the femoral head in hips with Legg–Calvé–Perthes disease. A decreased superior acetabular coverage in patients with Legg–Calvé–Perthes disease has been reported [5, 10] with larger values of Sharp's angle [37]. Global decrease in acetabular coverage has been shown in adult hips with Perthes disease, including dysplastic values for LCEA, IA and decreased anterior and posterior coverage of the femoral head [36]. In piglet models of Legg–Calvé–Perthes disease [38], a global decrease of the acetabular coverage was reported soon after ischemic osteonecrosis of the femoral head. In our study, we found that healed Legg–Calvé–Perthes disease hips are shallower as assessed by lower coronal ADR compared to the control hips. A decreased ADR has been previously found at all stages of Legg–Calvé–Perthes disease [10], potentially because the acetabulum deforms with greater width and smaller depth [5, 7, 9]. Changes in acetabular width and depth were more pronounced within the first year of the disease, suggesting that a partial acetabular remodeling occurs up to 5 years of follow-up [10].

In this study, cranial acetabular retroversion was observed in hips with healed Legg–Calvé–Perthes disease in comparison to the matched control hips. In contrast, a near normal cranial acetabular version was reported in adult hips with Legg–Calvé–Perthes disease [36]. Although the central acetabular version in our study was significantly lower in Legg–Calvé–Perthes disease hips, we

**Table IV. Comparison of acetabular morphology between the Legg–Calvé–Perthes characteristics**

| | *Stulberg* et al. *classification*[a] | | |
|---|---|---|---|
| *Acetabular measurements* | *II–III (14 hips)* | *IV–V (23 hips)* | P-*value* |
| Lateral center-edge angle (°) | 12.9±11.0 | 13.4±10.7 | 0.89 |
| Acetabular inclination angle (°) | 12.0±7.4 | 11.3±6.4 | 0.76 |
| Acetabular depth-width ratio | 291±43 | 285±47 | 0.69 |
| Anterior acetabular sector angle (°) | 52.3±9.0 | 52.5±10.0 | 0.94 |
| Posterior acetabular sector angle (°) | 81.1±5.9 | 78.3±5.8 | 0.15 |
| Cranial acetabular version (°) | 0.4±9.8 | 0.4±9.1 | 0.99 |
| Central acetabular version (°) | 15.2±3.8 | 12.8±5.6 | 0.17 |

[a]Acetabular measurements in hips with Legg–Calvé–Perthes disease (LCPD, $n = 37$ hips) summarized by mean and standard deviation and compared with the two-sample *t*-test. Cranial acetabular version was measured at a level 10 mm caudal to the acetabular dome, and central acetabular version at the level of the center of the femoral heads or Visser's centers in non-concentric or non-spherical hips.

believe that the mean central acetabular version of 13.7° was close to normative values [39]. Indeed, a difference of 3.4° compared with control hips may not be clinically important. In line with our findings, no differences were observed in the central acetabular version between skeletally immature Perthes hips and the contralateral unaffected hip [6]. Children with a more deformed femoral head may have the potential to develop acetabular retroversion over time, suggesting that the acetabular retroversion is an acquired deformity in Legg–Calvé–Perthes disease [6]. The phenomenon of acetabular retroversion in Legg–Calvé–Perthes disease, although well-described, is not fully understood [4, 6, 11, 14, 15, 36], and the retroversion may not be essentially a periacetabular phenomenon [14]. A study reported that the ischial spine sign was highly correlated with the presence of the crossover sign [14] and that this phenomenon would be associated with the torsion of the inferior hemipelvis [40]. Following this hypothesis, in cases of Legg–Calvé–Perthes disease, increased anterior tilt of the pelvis, combined with retroversion of the acetabulum and torsion of the hemipelvis, would be necessary during skeletal growth to maintain coverage of the deformed femoral head [14]. In piglet models of ischemic osteonecrosis of the femoral head, the acetabulum developed retroversion and decreased tilt, following the onset of femoral deformities [38]. Using 3D cartilage reconstruction from magnetic resonance imaging, a deformity of cartilage shape has been shown in Legg–Calvé–Perthes disease, and the distortion of the femoral surface preceded the acetabular surface deformity [12]. Nevertheless, further studies will be necessary to determine whether acetabular retroversion plays a role in the etiology of Legg–Calvé–Perthes disease [13, 41] or if it is an adaptive response to the deformation of the femoral head.

Our findings of a global acetabular insufficiency and cranial acetabular retroversion may assist the evaluation and treatment of hips with symptomatic healed Legg–Calvé–Perthes disease. Understanding the morphology of the acetabulum is critical to plan for treatment, both in the early stage of the disease and later, following the healing phase when the hip may become symptomatic [4, 20]. In early stages, improving the acetabular coverage of the femoral head by abduction bracing, femoral varus osteotomy or pelvic osteotomy has been the mainstay of Legg–Calvé–Perthes disease treatment (the containment principle). In the healed Legg–Calvé–Perthes disease hip, the complex 3D deformity may lead to structural instability or femoroacetabular impingement [4, 15, 20, 42]. Previous studies have recommended the triple [43] or the Bernese periacetabular osteotomy [20, 22, 42, 44] for correction of acetabular insufficiency in symptomatic hips. Although pelvic osteotomies may improve acetabular coverage of the femoral head [45], the surgeon should carefully assess the correction to avoid worsening the acetabular retroversion [46, 47].

We acknowledge several limitations of our study. First, our cohort of patients with Legg–Calvé–Perthes disease includes individuals who underwent CT scan for investigation of hip symptoms raising concern for selection bias. These patients may not represent the entire population with Legg–Calvé–Perthes disease, and our findings must be interpreted with caution due to their imperfect generalizability. It is possible that patients with continued symptoms and more deformed hips underwent complementary imaging investigation, while asymptomatic patients with more spherical hips were lost to follow-up or did not undergo CT scan evaluation. This bias can be anticipated by the low proportion of spherical hips (Stulberg *et al.* II) versus a greater proportion of aspherical hips (Stulberg *et al.* III, IV and V) in this series. Second, determining the femoral head center in aspherical hips is challenging, raising a concern for measurement bias. As a way to control this bias, we used the Visser's centers [29] for aspherical hips, and our reliability among the observers was satisfactory. Third, Legg–Calvé–Perthes disease may be associated with abnormalities in femoral version, including increased anteversion [36] and functional retroversion [48]. In the present study, we did not include the femoral version analysis, but the role of femoral version in the acetabular morphology warrants further investigation. Finally, although the assessors of measurements were not involved in the clinical care of patients, it was not possible to fully blind them to clinical characteristics because Legg–Calvé–Perthes disease deformities are evident in the CT images.

The acetabular morphology of skeletally mature hips with healed Legg–Calvé–Perthes disease is associated with shallow acetabulum, globally reduced coverage of the femoral head, and cranial acetabular retroversion in comparison with healthy hips. Our study helps to clarify the complex acetabular morphology and may be used as a reference for further research and when surgical treatment is considered for healed Legg–Calvé–Perthes disease.

## FUNDING

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## CONFLICT OF INTEREST STATEMENT

The other authors (M.F., L.A.K., W.H. and E.N.N.) have no conflict of interest to declare.

## ETHICAL STATEMENT

Ethical approval: This study was approved by our ethics and institutional review board committee.

Informed consent: Informed consent was not obtained from the patients included in this study given the retrospective design of the study.

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