

Asymmetrically increased femoral version with high prevalence of moderate and severe femoral anteversion in unilateral Legg-Calvé-Perthes disease

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Abstract

Purpose To determine and stratify femoral version in Legg-Calvé-Perthes disease (LCPD), and to compare the femoral version between the LCPD hip and the contralateral unaffected hip.

Methods We performed a retrospective review of 45 patients with unilateral LCPD who had available CT scan through the hips and knees between January 2000 and June 2017. There were 34 (76%) male cases with a mean age of 14 years (sd 4.69). Two independent readers measured femoral version on the affected and the unaffected contralateral femur. Femoral version was classified as follows: severely decreased version ($< 10^\circ$); moderately decreased (10° to 14°); normal femoral version range (15° to 20°); moderately increased (21° to 25°); and severely increased version ($> 25^\circ$).

Results LCPD hips had predominantly increased femoral version (38% severely increased anteversion, 24% moderately increased anteversion), while 51% of the contralateral unaffected hips had normal femoral version ($p < 0.001$). LCPD hips had higher mean femoral version than the contralateral, unaffected side (mean difference = 13° ; 95% confidence interval 10° to 16° ; $p < 0.001$). As the version of the affected hip increased, so did the discrepancy between sides. No effect of sex on the LCPD femoral version was detected ($p = 0.34$).

Conclusion This study included a selected group of patients with unilateral LCPD and available CT scans obtained for sur-

gical planning. The femoral version was asymmetric, with a high proportion of excessive anteversion observed at later stages of disease in the affected hips. Future studies will be necessary to determine the pathogenesis of increased femoral version associated with LCPD.

Level of Evidence: Level IV, retrospective study.

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Introduction

Legg-Calvé-Perthes disease (LCPD) is a paediatric hip disorder characterized by idiopathic osteonecrosis of the immature capital femoral epiphysis.¹ Prevention of severe femoral head deformity and joint incongruity is the primary goal of treatment.²⁻⁴ In the most severe cases, a flat or mushroom-shaped femoral head incongruous with the acetabulum may develop, leading to hip pain from femoroacetabular impingement (FAI), instability, chondral and labral disease and early osteoarthritis.^{5,6} The residual deformity of the proximal femur, including a large femoral head with a short femoral neck and a high riding greater trochanter, has been well described in the literature. However, the characterization of the torsional deformity of the femur in LCPD remains controversial.⁷⁻¹³ Some studies reported normal or only mildly increased femoral version in LCPD patients relative to healthy controls, with a low frequency of increased femoral version.^{10,11,13} In contrast, others reported excessive femoral version in patients with LCPD.^{7-9,12} However, most previous studies have investigated the femoral version with plain radiographs instead of contemporary methods, including CT or MRI.

The purpose of this study was: 1) to determine the femoral version in the affected femur of patients with unilateral LCPD; and 2) to compare the femoral version in the affected *versus* the contralateral uninvolved femur by using axial CT of the pelvis extending to the distal femur.

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Materials and methods

Study design

Our institutional review board approved this retrospective study. We searched our institutional database for patients diagnosed with LCPD between 1st January 2000 and 30th June 2017. The inclusion criteria were: 1) diagnosis of unilateral LCPD as confirmed by a history of limp or hip pain, and radiographs; and 2) an available CT scan through the hips and knees before any surgical intervention. During the study period, the indication for CT scan was to help with surgical treatment planning of a hip deformity secondary to LCPD related to hinge abduction, FAI or hip instability. The initial search identified 496 patients treated for LCPD in our institution during the study period. However 392 (79%) were excluded because they did not undergo a CT scan. After review of the medical records and imaging 59 out of 104 (57%) patients were excluded yielding a total of 45 patients (45 hips) included in the study (Fig. 1). There were 34 (76%) male cases, and the mean age at the time of CT was 14 years (SD 5; 6 to 26). The triradiate cartilage was open in 19 (42%) patients and it was closed in 26 (58%) patients. The CT was performed after a median of 3.6 years (interquartile range 1.3 to 8; 0 to 17) after the diagnosis of LCPD. All hips were categorized according to the modified Waldenström classification,¹⁴ using radiographs obtained at the same time as the CT acquisition. In total, 38 out of the 45 hips (84%) were considered in the residual stage of the disease; three (7%) hips were in

the reossification stage and four (9%) hips were in the late fragmentation stage.

CT imaging and assessment of femoral version

For CT acquisition, the patients were positioned supine with two small bean bags supporting the lateral aspect of the leg to hold the patella in a neutral position. Axial CT was performed, including the pelvis and the distal femoral condyles on a 1 mm slice thickness. One of the authors (MGF) who was not involved in the clinical care of the patients measured the femoral version in all 45 patients (90 hips) following the technique described by Weiner et al.¹⁵ An axial image of the proximal femur in which the anterior and posterior cortices were parallel to each other was selected. The femoral neck axis was defined as the midline between the anterior and posterior cortices. The femoral neck angle was measured as the angle formed by the femoral neck axis and a horizontal line. Distally, the axis of the femoral condyles was defined as the posterior tangent line to the femoral condyles. The distal femoral condyle angle was measured as the angle formed by the posterior condylar line and a horizontal line. The femoral version angle was calculated based on the femoral neck angle and the distal femoral condyle angle. If the posterior condylar line was rotated outward relative to the horizontal line, then the distal femoral condyle angle was subtracted from the femoral neck angle. If the posterior condylar line was rotated inward relative to the horizontal line, then the distal femoral condyle angle was added to the femoral neck

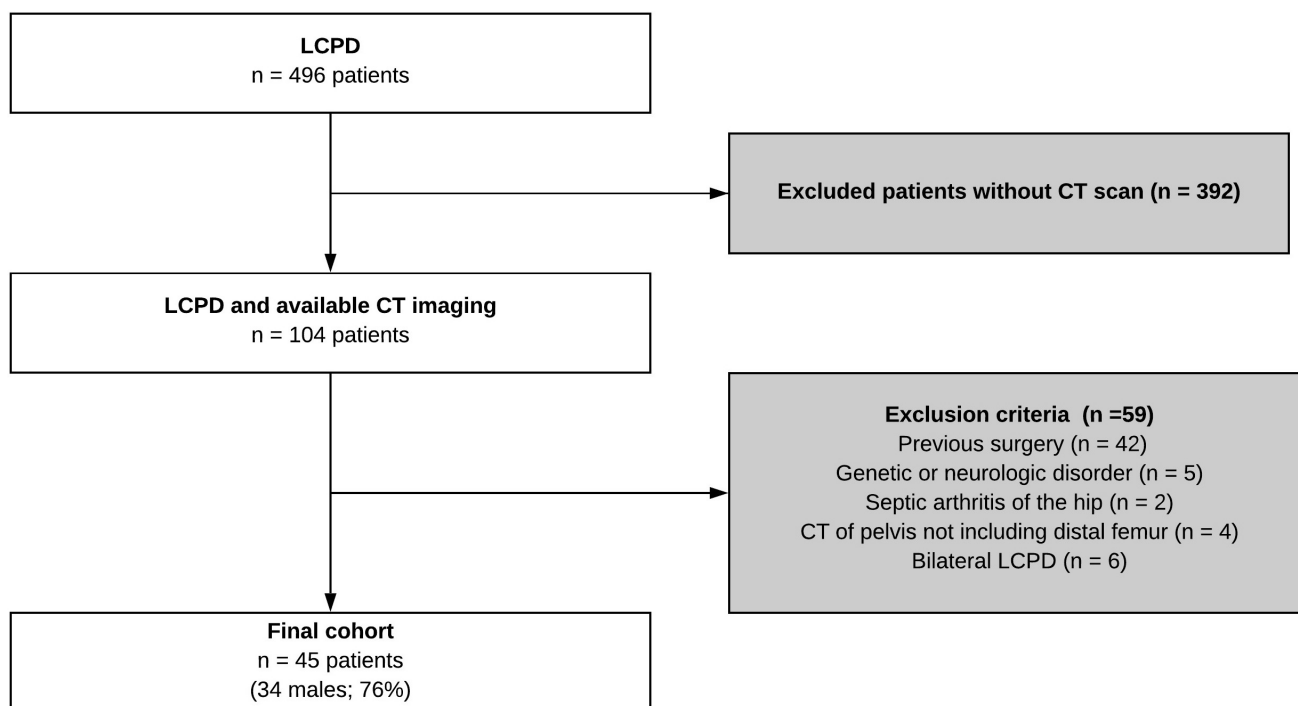


Fig. 1 Diagram showing the selection criteria for the study cohort (LCPD, Legg-Calvé-Perthes disease).

angle (Fig. 2). The calculated femoral version was then classified according to the categories described by Tönnis and Heinecke¹⁶ as follows: severely decreased version ($< 10^\circ$); moderately decreased (10° to 14°); normal femoral version range (15° to 20°); moderately increased (21° to 25°); and severely increased version ($> 25^\circ$).

A random selection of 19 CT scans was reviewed by two independent reviewer (MGF,JDW) at a separate time approximately six weeks after the first reading. The femoral version of the affected and contralateral unaffected hip was measured in each case. For intrarater reliability an intraclass correlation coefficient (ICC) model was used which is a two-way mixed effects model for a single rater to assess consistency. For interrater reliability an ICC model was used which is a two-way random effects model

using the average of two random raters to assess agreement. Intra- and interrater reliability of femoral version measurements was excellent for the affected side (ICC = 0.997 and 0.994, respectively) and the contralateral side (ICC = 0.996 and 0.994, respectively).

Statistical analysis

Patient characteristics and femoral version were summarized for all patients by frequency and percentage, or mean and SD, as appropriate. The femoral version was compared between affected and contralateral hips using general linear modelling with a generalized estimating equations approach to control for clustering within each patient. The least-squares means were computed for

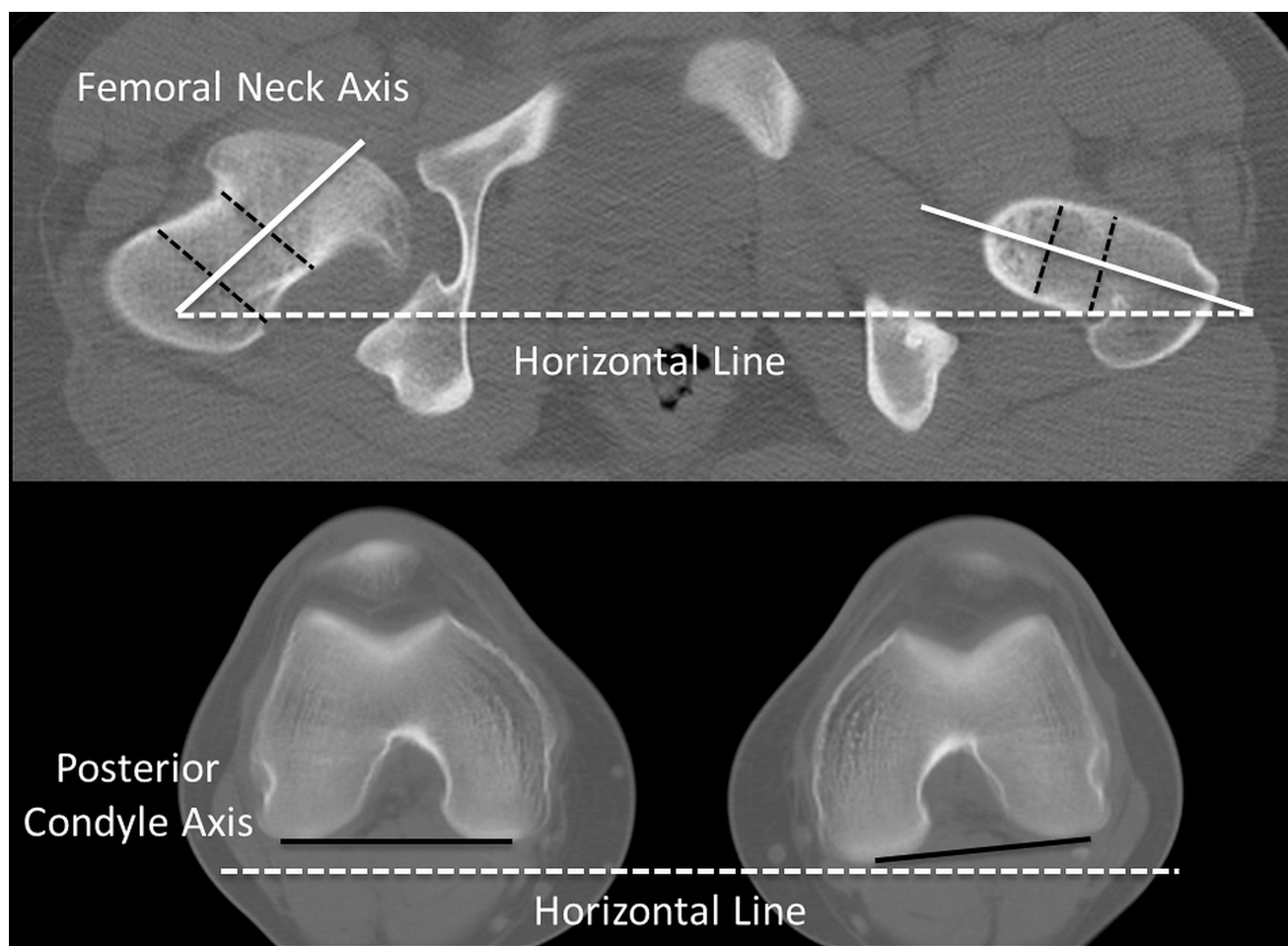


Fig. 2 Measurement of femoral version based on axial CT image through the proximal and distal femur. An axial image of the proximal femur in which the anterior and posterior cortices were parallel to each other is used to draw the femoral neck axis (white solid line) defined as the midline between the anterior and posterior cortices (assessed by the two dashed black lines). The femoral neck angle was measured as the angle formed by the femoral neck axis and the horizontal line. At the level of the knee, the axis of the femoral condyles was defined as the posterior tangent line to the femoral condyles. The distal femoral condyle angle was measured as the angle formed by the posterior condylar line and a horizontal line. The femoral version angle was calculated based on the femoral neck angle and the distal femoral condyle angle. If the distal femur was rotated outward relative to the proximal femur, the distal femoral condyle angle was subtracted from the femoral neck angle. If the distal femur was rotated inward relative to the proximal femur, the distal femoral condyle angle was added to the femoral neck angle.

affected and contralateral sides and stratified by version severity and were adjusted using Bonferoni's adjustment for multiple comparisons. The mean difference and corresponding adjusted 95% confidence intervals (CIs) were reported for differences between affected and contralateral sides. General linear modelling was used to determine if there was an effect of sex on femoral version measurement on the affected LCPD hip. All tests were two-sided, and a p-values < 0.05 was considered significant.

Power analysis determined that a sample of 45 subjects would provide 80% power to detect differences in femoral version of at least 6° between affected and contralateral hips. This was based on conducting an independent samples Student's *t*-test with alpha set to 5%. The final analysis accounted for the correlation between measurements on the same patient and expectedly provided estimates with a higher power than the test used in the *a priori* power analysis.

Results

LCPD hips had predominantly increased femoral version (38% severely increased anteversion, 24% moderately increased anteversion), while 51% of the contralateral unaffected hips had normal femoral version ($p < 0.001$)

(Fig. 3). LCPD hips had higher mean femoral version than the contralateral unaffected hip (mean difference 13°; 95% CI 10° to 16°; $p < 0.001$). Overall, as the femoral version of the LCPD hips increased, the difference between the femoral version of the affected and the unaffected contralateral side also increased. Adjusted comparisons indicated that patients with severely increased femoral version on the affected LCPD hip had a significantly lower mean femoral version on the contralateral side (mean difference 18°; 95% CI 12° to 25°; $p < 0.001$) (Table 1). Similarly, patients with moderately increased femoral version on the affected hip had a significantly lower mean femoral version on the contralateral side (mean difference 15°; 95% CI 4° to 25°; $p < 0.001$). Patients with moderately decreased femoral version on the affected hip had a significantly lower mean femoral version on the contralateral side (mean difference 7°; 95% CI 1° to 13°; $p = 0.004$). However, we detected no statistically significant difference between the LCPD and the unaffected contralateral side for patients with severely decreased femoral version; $p = 0.99$).

There was no effect of sex detected on the femoral version ($p = 0.34$) for the entire cohort or across severity groups (Table 2). Furthermore, we found no association ($p = 0.12$) between the length of time between diagnosis and CT scan and severity of increased femoral version.

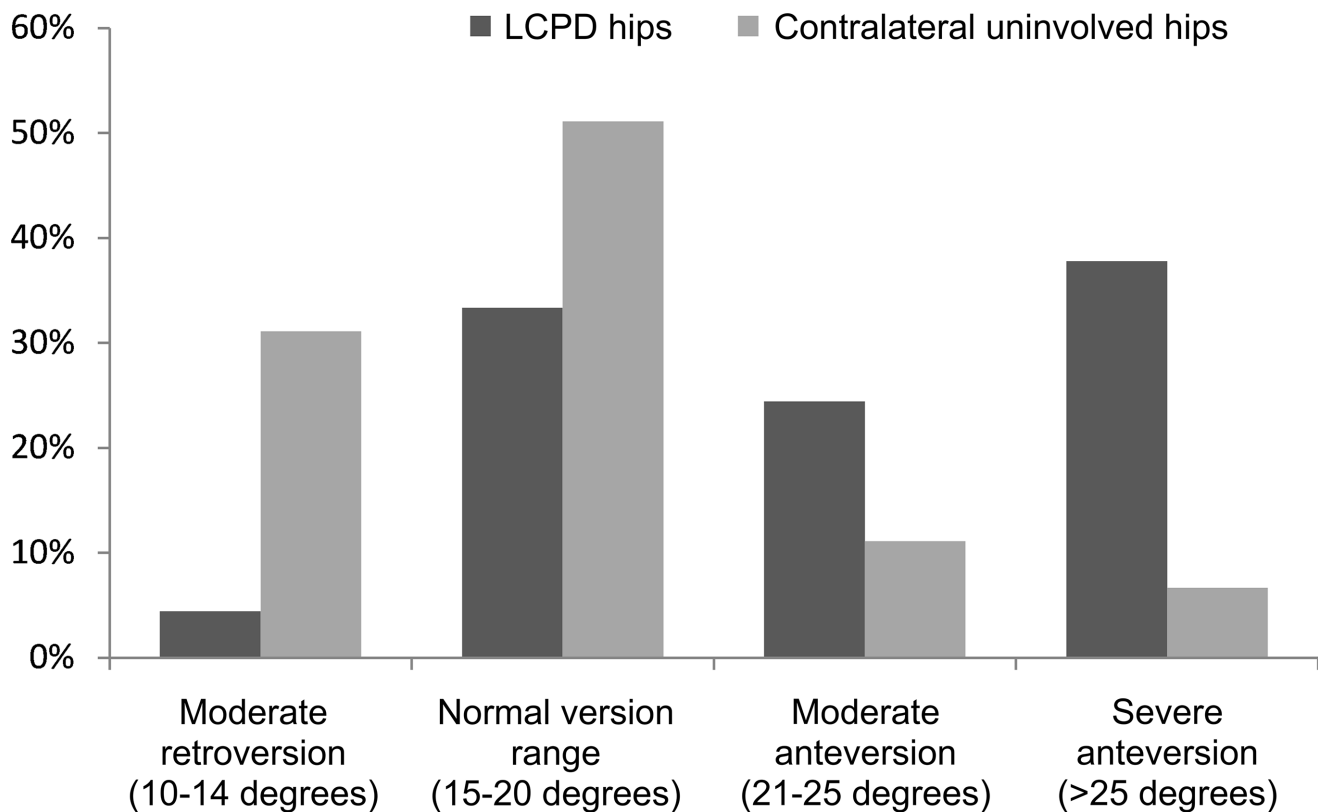


Fig. 3 Diagram showing the distribution of femoral version according to the Tonnis and Heinecke¹⁶ criteria between Legg-Calvé-Perthes disease (LCPD) hips and the contralateral uninvolved hip.

Table 1 Comparisons between Legg-Calvé-Perthes disease (LCPD) and contralateral uninvolved hips by affected side version severity (n = 45). Least-squares means and confidence intervals are reported based on a general linear model using a general estimating approach for clustering within the same patient

Femoral version	n (%)	LCPD	Contralateral	p-value*
		Mean, ° (95% CI)	Mean, ° (95% CI)	
All data	45 (100)	29.2 (26.2 to 32.1)	16.3 (13.3 to 19.3)	< 0.001
Severe retroversion	0 (0)	0.0	0.0	-
Moderate retroversion	2 (4)	4.0 (3.3 to 4.8)	3.8 (-1.4 to 9.1)	0.99
Normal version	15 (33)	18.3 (16.1 to 20.4)	11.3 (8.0 to 14.6)	0.004
Moderate anteversion	11 (24)	30.9 (29.5 to 32.2)	16.2 (9.6 to 22.7)	< 0.001
Severe anteversion	17 (38)	40.7 (38.5 to 42.8)	22.4 (18.3 to 26.5)	< 0.001

*pairwise comparison p-values have been adjusted using a Bonferonni correction
CI, confidence interval

Discussion

Assessment of the femoral version in patients with LCPD is essential for deformity analysis and surgical planning. To the best of our knowledge, torsional deformity in LCPD has not been fully described. In this study, we measured the femoral version in 45 patients with LCPD and stratified the severity according to a previously described classification system.¹⁶ We found that 62% of patients with LCPD had moderately or severely increased femoral version, 34% had a normal version, and 4% had moderately decreased version. In contrast, the majority of the contralateral hips had normal or moderately decreased femoral version. LCPD hips had higher mean femoral version than the contralateral unaffected hip. The higher the anteversion in the affected hip, the higher the discrepancy between the two hips.

Most previous studies evaluating the femoral version in LCPD utilized plain films and reported conflicting results as far as the distribution of the femoral version of the affected and unaffected hips.^{7-11,13} Some studies reported a high prevalence of increased femoral version in LCPD. Dunlap et al⁹ found an increased femoral version in 60% of hips in 25 patients with LCPD. Craig et al⁸ found that the average anteversion was 45°, and several patients had as high as 60° of anteversion. Axer et al⁷ measured the femoral version using radiographs in 68 patients with LCPD and found that 53% of LCPD hips had 10° or more of femoral version when compared with normal hips. On the contrary, other studies reported a low prevalence of

an increased femoral version in LCPD. Katz¹¹ reported that only 20% (11/54) of LCPD hips had a femoral version of 10° or more above the normal average for the age range in 49 patients (54 hips) with LCPD. Shands and Steele¹³ reported that 15% of LCPD hips had a femoral version of 10° or more above the normal average for the age range. Similarly, Fabry¹⁰ reported that 22% of the LCPD hips had a femoral version of 10° or more above the normal. Our data suggest the femoral version is typically increased in LCPD. In our study, 62% of patients with LCPD had increased femoral version. Our findings are in line with those reported by Lerch et al¹² who reported that 50% of patients with LCPD had severely increased femoral version assessed by CT or MRI.

Our findings are merely observational, and we cannot infer whether excessive femoral anteversion is a precipitating factor for the development of LCPD, or if it is a consequence of the disease process. However, the contralateral femur was found to have an increased femoral version in only 15% of the patients with LCPD. Similarly, Axer et al⁷ found that 17.5% of the contralateral hips had 10° or more of the femoral version compared with normal hips. The discrepancy between the affected and unaffected side suggests that excessive femoral anteversion develops as a consequence of the osteonecrosis, remodelling and further asymmetric growth associated with LCPD. The shape of the proximal femoral growth plate influences the femoral version in normal development with gait loading reducing femoral anteversion with growth.¹⁷ One study suggested that growth disturbance of the femur

Table 2 Legg-Calvé-Perthes disease affected hip femoral version summary by version severity and sex (n = 45)

Femoral version	Female		Male		p-value*
	n (%)	Mean, ° (sd)	n (%)	Mean, ° (sd)	
All data	11 (24)	25.4 (15.48)	34 (76)	30.4 (10.17)	0.34
Severe retroversion	0	-	0	-	-
Moderate retroversion	1 (9)	4.6 (-)	1 (3)	3.5 (-)	-
Normal version	6 (55)	17.2 (5.24)	9 (26)	18.9 (3.89)	1.00
Moderate anteversion	0 (0)	-	11 (32)	30.9 (2.47)	-
Severe anteversion	4 (36)	42.9 (8.04)	13 (38)	40.0 (3.39)	1.00

*pairwise comparison p-values have been adjusted using a Bonferonni correction

secondary to the ischemic damage may lead to a lack of spontaneous correction of anteversion that is expected with growth.¹⁸ Although the increased femoral version observed in our study may be related to changes in the direction of growth associated with the physal growth arrest, further research is needed to determine the etiopathogenesis of increased femoral version in hips with LCPD. Complete characterization of the femoral version will require a large population of patients with LCPD with a cross-sectional image of the pelvis and knee at the time of diagnosis and during follow-up.

Assessment of the femoral version is essential to help understand the aetiology of hip pain in patients with healing ossification of the femoral head or in those with residual deformity after LCPD and for accurate surgical planning before a femoral osteotomy. Excessive femoral version aggravates the hip instability and the extraarticular impingement between the greater trochanter and the posterior acetabulum and ischium.¹⁹ On the other side of the spectrum, femoral retroversion increases the risk of intraarticular FAI.^{5,20-22} Although treatment of LCPD remains highly controversial, femoral intertrochanteric varus osteotomy is a widely accepted option during the early phase of the disease.^{23,24} A femoral valgus osteotomy is one of the surgical options for the treatment of hinge abduction.^{25,26} A derotational osteotomy has been recommended to improve intraoperative hip instability during a surgical hip dislocation with osteochondroplasty of the femoral head and relative neck lengthening.²⁷ Although our data suggest that the femoral version is typically increased in patients with healed LCPD, there was a variation in measurements that should be considered. Therefore, we do not recommend routine correction of excessive femoral version by derotation of the femur in patients undergoing femoral osteotomy for the treatment of LCPD in the acute onset or during later reconstruction. Instead, we recommend cross-section imaging for preoperative assessment of the femoral version as well as intraoperative evaluation of the hip range of movement in flexion and extension.

Our study has some limitations. First, because of the relatively small number of patients in our study, we could not fully investigate whether the degree of femoral version differed by different age groups. Hence, we can infer little concerning the interplay between age and femoral version in patients with LCPD. In the healthily developing child, the degree of femoral version decreases with increasing age.¹⁶ However, Axer et al⁷ found that in LCPD, femoral anteversion does not decrease with increasing age. As such, those authors concluded that excessive femoral anteversion in LCPD hips is more likely a result of the disease process, not a causative factor for the disease. Second, the retrospective design of the study is asso-

ciated with a potential for selection bias as the patients included in the study do not represent the entire population of LCPD patients seen at our institution during the study period. In this study, the indication for CT scan was to help with surgical treatment planning of a hip deformity secondary to LCPD related to hinge abduction, FAI or hip instability. Therefore, it is possible that the patients included in this study would have a more severe deformity than the general patients with LCPD. This complication is difficult to overcome with the retrospective nature of the study. A prospective study is needed to investigate the development of rotational deformity of the femur associated with LCPD. Third, we did not consider the potential interaction between the femoral version and severity of LCPD because CT was obtained at a late stage with healed LCPD deformity, and we did not have information about the involvement of femoral head at the time of LCPD onset in every patient. Fourth, we made our observations made on the axis of the femoral neck, therefore, not accounting for the potential functional retroversion of the deformed femoral head.²⁸

In conclusion, our study found a high prevalence of increased femoral version in patients with LCPD. However, the uninvolved contralateral hip had a typically normal femoral version. This side-to-side discrepancy is a gradient that appears to increase as the femoral version increases in hips with LCPD. Our findings should be considered when planning a femoral osteotomy for the treatment of patients with LCPD undergoing surgical reconstruction. Preoperative cross-section imaging and intraoperative assessment may optimize the planning and performance of a femoral osteotomy to avoid inadvertent changes of the femoral version that can aggravate hip instability or FAI.

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COMPLIANCE WITH ETHICAL STANDARDS

FUNDING STATEMENT

No benefits in any form have been received or will be received from a commercial party related directly or indirectly to the subject of this article.

OA LICENCE TEXT

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ETHICAL STATEMENT

Ethical approval: All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. The study was approved as a retrospective study by our institutional review board.

Informed consent: Informed consent was not obtained from subjects in this retrospective study.

ICMJE CONFLICT OF INTEREST STATEMENT

James D. Wylie reports research funding from Arthrex, Inc., is an editorial board member for Arthroscopy and a board committee member for AOSSM

AUTHOR CONTRIBUTIONS

ENN: Final approval of the version to be published, Drafting the work or revising it critically for important intellectual content, Interpretation of data for the work.

KDN: Drafting the work or revising it critically for important intellectual content, Final approval of the version to be published.

MGF: Final approval of the version to be published, Conception or design of the work, Acquisition, analysis and interpretation of data for the work.

PEM: Drafting the work or revising it critically for important intellectual content, Analysis or interpretation of data for the work.

JDW: Final approval of the version to be published, Conception or design of the work, Acquisition, analysis and interpretation of data for the work.

WTD: Interpretation of data for the work, Drafting the work or revising it critically for important intellectual content.

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