ORIGINAL ARTICLE

Ipsilateral Osteochondritis Dissecans-like Distal Femoral Lesions in Children with Blount Disease: Prevalence and Associated Findings

Folorunsho Edobor-Osula[1], Cornelia Wenokor[2], Tamir Bloom[3], Caixia Zhao[4], Sanjeev Sabharwal[5]

Abstract

Purpose: Our goal was to assess the prevalence of ipsilateral distal femoral osteochondritis dissecans (OCD)-like lesions in children with Blount disease, including factors associated with this finding.

Materials and methods: Characteristics of patients with an OCD-like lesion on an imaging study [(X-ray and/or magnetic resonance imaging (MRI)] were compared with those without such a finding.

Results: Over a 12-year period, 6/63 (10%) skeletally immature patients (9/87 limbs) with Blount disease had an OCD-like lesion visible on plain radiographs. Based on available MRI, 7/37 (19%) patients or 10/53 (19%) limbs had an OCD-like distal femoral lesion. All lesions were noted in the posterior third of the weight-bearing portion of the medial femoral condyle with intact overlying articular cartilage. All patients with OCD-like lesions were followed for an average of 1.9 years (range: 1–2.6 years), and complete radiographic resolution of lesion was noted in 7/9 limbs (78%). There was no association of the presence of OCD-like lesion with early- vs late-onset disease, gender, age at imaging, laterality, magnitude of deformity [mean mechanical axis deviation (MAD) 63.3 vs 71.9 mm], mean mechanical lateral distal femoral angle (mLDFA; 91.3 vs 89.7°), and mean medial proximal tibial angle (MPTA; 71.7 vs 71.8°). Children with an OCD-like lesion tended to have a lower mean body mass index (BMI; 21 vs 36, $p = 0.003$).

Conclusion: The overall prevalence of OCD-like lesions in the medial femoral condyle in children with Blount disease lesions is 10% using plain radiographs and at least 19% on MRI. Based on the numbers available, we were unable to demonstrate any associations between the presence of such lesions and the patient's age, gender, or magnitude of varus deformity. Further research is needed to fully ascertain the aetiology and natural history of these benign appearing osteochondral imaging findings in children with Blount disease. Our current data support that these lesions do resolve with time and that no surgical intervention targeted at the femoral OCD-like lesion is warranted.

Level of evidence: Diagnostic study Level III.

Keyword: Blount disease, genu Varum, Irregular ossification, Osteochondritis Dissecans.

Strategies in Trauma and Limb Reconstruction (2019): 10.5005/jp-journals-10080-1438

Introduction

Blount disease or tibia vara affects the posteromedial aspect of the proximal tibia in children causing a bowing deformity with procurvatum of the lower extremity.[1] Two clinically distinct types exist based on the age at which the deformity occurs, early onset (occurring before age 4) and late onset (occurring after age 4).[2] Assessment of Blount disease is primarily based on radiographic findings. However, with the advent of magnetic resonance imaging (MRI), the intra-articular morphology can be evaluated in greater detail.[3] Over the years, we have encountered several skeletally immature patients with Blount disease who have demonstrable osteochondral findings in the ipsilateral medial distal femoral epiphysis that closely resemble osteochondritis dissecans (OCD)-like lesions on plain radiographs and MRI.

The purpose of our study was to evaluate the prevalence of ipsilateral distal femoral OCD-like lesions in children with Blount disease. Additionally, we planned to describe the morphologic features of these lesions based on plain radiographs and MRI (when available) and evaluate any clinical factors that may be associated with such radiologic findings.

Materials and Methods

After institutional review board approval, the medical records of all patients with a diagnosis of Blount disease treated between January 2005 and March 2016 at a single institution were retrospectively reviewed, using a paediatric orthopaedic database. Exclusion criteria included patients with poor quality or incomplete plain X-rays, incorrect diagnosis code, and patients who were adults (with closed growth plates around the knee) at initial presentation. All patients

[1,4]Department of Orthopedics, Rutgers-New Jersey Medical School, New Jersey, USA

[2]Department of Radiology, Rutgers-New Jersey Medical School, New Jersey, USA

[3]The Pediatric Orthopedic Center, Cedar Knolls, New Jersey, USA

[5]Department of Orthopedic Surgery, University of California, San Francisco, USA; UCSF Benioff Children's Hospital, Oakland, California, USA

Corresponding Author: Sanjeev Sabharwal, Department of Orthopedic Surgery, University of California, San Francisco, USA; UCSF Benioff Children's Hospital, Oakland, California, USA, e-mail: Sanjeev.sabharwal@ucsf.edu

How to cite this article: Edobor-Osula F, Wenokor C, Bloom T, *et al.* Ipsilateral Osteochondritis Dissecans-like Distal Femoral Lesions in Children with Blount Disease: Prevalence and Associated Findings. Strategies Trauma Limb Reconstr 2019;14(3):121–125.

Source of support: Nil

Conflict of interest: None

included in this study had a preoperative standing full-length anteroposterior (AP) radiograph of both lower extremities and AP and lateral views of the involved knee. When available, a preoperative MRI was reviewed using a Signa 1.5-T systems (General Electric Medical System, Milwaukee, WI) with a transmit–receive knee coil.

Demographic information was extracted including patient's age at time of diagnosis and imaging, gender, BMI, and affected side(s). Each included patient's radiographs and MRI (if available) were reviewed for evidence of OCD-like lesion in the ipsilateral distal femur with Blount disease. All patients noted to have an OCD-like lesion were identified, and each such imaging study was reviewed by three independent examiners, a musculoskeletal radiologist (C.W.) and two paediatric orthopaedic surgeons (F.O. and T.B.). Each patient's OCD-like lesion on MRI was graded according to two validated staging systems used to evaluate OCD lesions. In the Dipaola system,[4] lesions are graded I–IV, depending on the breach of the overlying articular cartilage and the stability of the lesion based on increased fluid signal behind the fragment on T2 imaging. The Hefti staging system[5] evaluates the stability of the fragment based on increased fluid signal between the fragment and the underlying bone and is graded I–V.

The preoperative radiographs of all patients (with and without OCD-like lesions) were reviewed and data regarding the type of Blount disease: early-onset, defined as lower limb deformity occurring before age 4 years, or late-onset, if noted after age 4 years, was recorded. The lower limb deformity parameters were measured[1] including the mechanical axis deviation (MAD), mechanical lateral distal femoral angle (mLDFA), and mean medial proximal tibial angle (MPTA) by a fellowship trained paediatric orthopaedic surgeon (F.O.). Patients were followed postoperatively, and presence or absence of a lesion was noted on plain radiographs of the knee. Intra-observer reliability was calculated using a random set of 10 radiographs that were measured 1 month apart. The two groups (with and without OCD-like lesions) were compared for any differences in demographics and magnitude of radiographic deformities. Statistical analysis was performed, including Student's *t* test for comparison of continuous variables and Chi-square for categorical variables. The intra-observer and inter-observer reliability was measured using the intra-class correlation coefficient (ICC). An ICC of 0–0.24 reflects absent to poor, 0.25–0.49 low, 0.50–0.69 fair/moderate, 0.7–0.89 good, and 0.9–1.0 excellent correlation. Differences were considered statistically significant at $p < 0.05$.

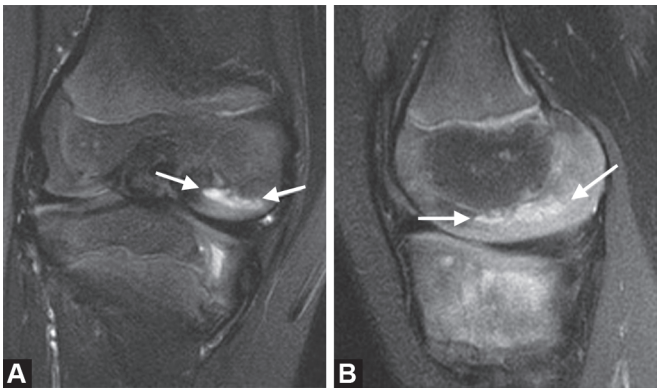
Results

A total of 68 patients with Blount disease were identified. Five patients were excluded: two patients were disqualified due to inadequate imaging, and three patients were adults at initial presentation. Demographic features of the remaining 63 patients are summarised in Table 1.

All 63 patients (87 affected limbs) had plain radiographs, including full-length standing radiographs of the lower extremities and dedicated AP and lateral views of the affected knee. A total of nine OCD-like lesions (in six patients) were identified on plain radiographs, with an overall prevalence of 10% (9/87). From a total of 37 patients (53 limbs) who also had an MRI of the knee, 7 (19%) patients with 10/53 (19%) limbs had an OCD-like lesion noted on their MRI. All lesions were found in the posterior one third of the weight-bearing portion of the medial femoral condyle (Figs 1 to 3). The mean dimensions of the lesion were similar, based on plain radiographs vs MRI (Table 2). The measurement variability for plain

Table 1: Demographic Information on all patients with Blount disease

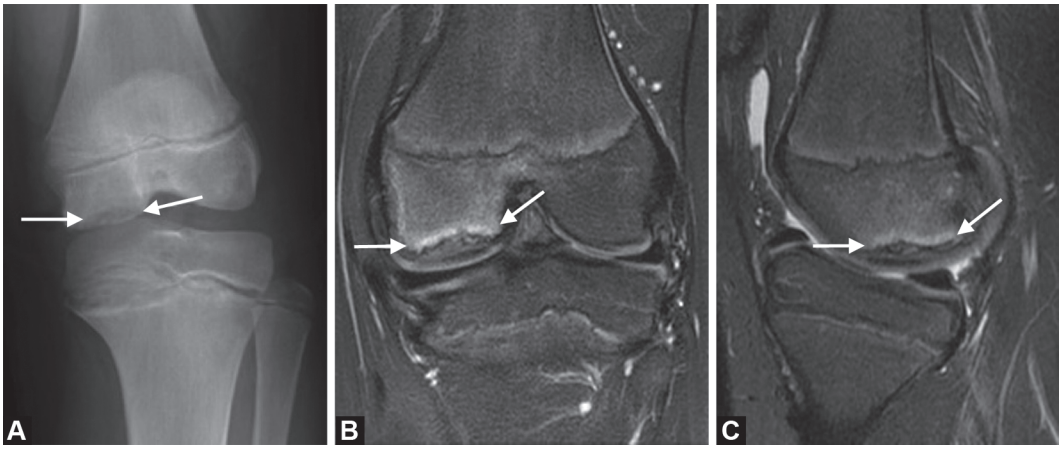
	Early-onset (%)	*Late-onset (%)*	*Total*
Patients [no. (%)]	21 (33)	42 (67)	63
Limbs [no. (%)]	36 (41)	51 (59)	87
Gender [no. (%)]			
Male	9 (43)	32 (76)	41 (65)
Female	12 (57)	10 (24)	22 (35)
Mean age at X-rays [year (range)]	7.2 (2–18.1)	12.6 (7.8–18.3)	10.8 (2–18.3)
Side [no. (%)]			
Right	1 (5)	15 (36)	16
Left	5 (24)	18 (43)	23
Bilateral	15 (71)	9 (21)	24



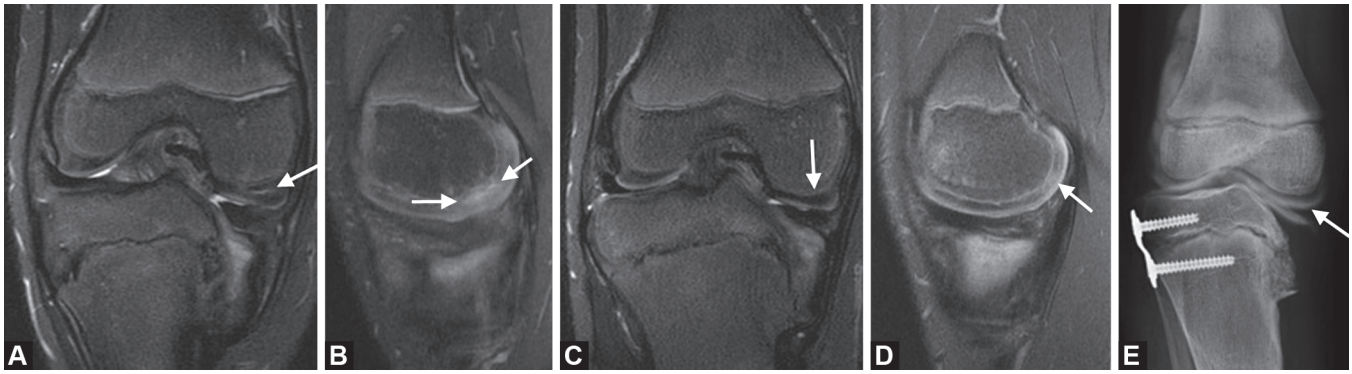
Figs 1A and B: A 3-year-old boy with early-onset Blount disease. Coronal (A) and sagittal (B) fat-suppressed T2-weighted images demonstrate a fluid-filled cleft (white arrows) in the hypertrophied medial femoral condylar cartilage. The surface of the articular cartilage is intact. There is slight medial meniscal hypertrophy

radiographic parameters was excellent (ICC: 0.81–0.96) and good to excellent (ICC: 0.77–0.86) for MRI findings. As expected, the size of the lesion was slightly larger in the late-onset group with the mean lesion size on MRI measuring 152.7 mm^2 in the early-onset group and 178.5 mm^2 in the late-onset group ($p = 0.064$). Using the Dipaola system, all lesions were classified as stage I, indicating no breach of the overlying distal femoral articular cartilage.[4] Based on the Hefti classification, there were three stage I, two stage II, and five stage III lesions.[5] The typical MRI features of the OCD-like femoral lesions included fluid-filled clefts either within the hypertrophied medial femoral condylar cartilage or at the bone–cartilage interphase with mild adjacent bone marrow oedema (Figs 1 to 3). Some patients also demonstrated cortical irregularity of the weight-bearing surface of the medial femoral condyle with adjacent marrow oedema (Fig. 2). These changes were best seen on fat-suppressed T2-weighted and proton density sequences.

There was no statistically significant difference between the groups (with and without OCD-like lesions) in terms of diagnosis: early-onset vs late-onset disease ($p = 0.21$), gender ($p = 0.23$), mean age at imaging ($p = 0.06$), and laterality ($p = 0.07$). Children with an OCD-like lesion tended to have a lower mean BMI (21 vs 36, $p = 0.003$) (Table 3). There was also no significant difference between the two groups in terms of mean MAD (63.3 mm vs 71.9 mm, $p = 0.39$), mean mLDFA (91.3° vs 89.7°, $p = 0.43$), and mean MPTA (71.7° vs 71.8°, $p = 0.95$) (Table 4). Patients were followed up



Figs 2A to C: A 10-year-old boy with late-onset Blount disease. Plain radiographs (A) of the left knee demonstrating an OCD-like lesion with an irregular-bordered crescentic lucency (white arrows) at the weight-bearing surface of the medial femoral condyle. Coronal (B) and sagittal (C) fat-suppressed T2-weighted images demonstrate cortical irregularity of the medial femoral condyle with adjacent marrow oedema (white arrows). The surface of the overlying articular cartilage is intact



Figs 3A to E: A 5-year-old boy with early-onset Blount disease. Coronal (A) and sagittal (B) fat-suppressed T2-weighted images demonstrate a small fluid-filled cleft (white arrows) in the hypertrophied medial femoral condylar cartilage. The surface of the articular cartilage is intact. There is marked medial meniscal hypertrophy. Follow-up 1 year later. Sagittal fat-suppressed T2-weighted images demonstrate resolution of the cleft with residual mild cartilage oedema on coronal (C) and sagittal (D) MRI images. Completely normal appearing cartilage on the coronal image (white arrows). Persistent hypertrophied medial femoral condylar cartilage and medial meniscal hypertrophy. Intraoperative arthrogram also demonstrates intact articular cartilage (white arrow) (E)

Table 2: Comparing dimensions of osteochondritis dissecans (OCD)-like lesions as measured on plain radiographs vs MRI

	Radiographs means (95% CI)	*MRI means (95% CI)*	*Mean difference*	*p value*
Lesion width (mm)	11.5 (8.9–14.1)	10.9 (8.3–13.6)	0.6	0.74
Lesion length (mm)	15.4 (12.8–18.0)	14.1 (11.5–16.8)	1.3	0.47
Lesion surface area (mm^2)	197.2 (133.9–260.5)	163.0 (107.6–218.5)	34.2	0.36

for an average of 1.9 years (range: 1–2.6 years) after identification of the OCD-like lesion on plain radiographs or on MRI, and a complete radiographic resolution of lesion was noted in 7/9 (78%) limbs (Fig. 4). Of the remaining two limbs, the lesion was noted to have decreased in size. No patient had a lesion that had remained the same size on follow-up imaging. None of the patients developed any mechanical symptoms or required surgical intervention for the OCD-like lesions.

Discussion

To our knowledge, this is the first study to report on the presence of OCD-like distal femoral lesions in children with Blount disease. We found that the overall prevalence of OCD-like lesions in the medial femoral condyle in children with Blount disease lesions was 10% using plain radiographs and at least 19% on MRI. While our study does not give conclusive evidence of the aetiology of these lesions, it does provide some useful information. First, long-term follow-up demonstrates that these lesions largely resolve over time and/or significantly decrease in size. None of the patients in our cohort required surgical intervention for these lesions. Second, all the lesions were found in the posterior one third of the weight-bearing portion of the medial femoral condyle. Anatomic studies have shown that this is an uncommon location for accessory ossification centers of the distal femur.[6,7] In a study by Caffey et al., anomalies of ossification were nearly four times more commonly seen in the posterior aspect of the lateral femoral condyle when compared with the medial femoral condyle.[8] These changes are oftentimes found serendipitously by the treating orthopaedic surgeon and

Table 3: Comparison of demographic information between patients with and without osteochondritis dissecans (OCD)-like lesions

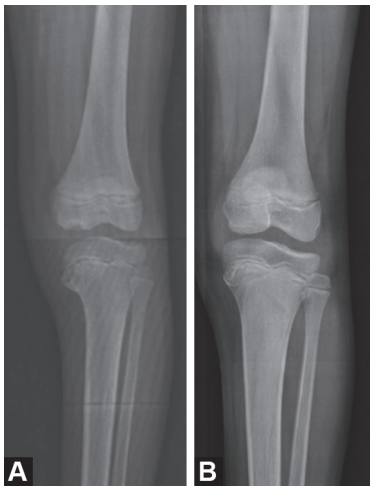
	OCD-like lesion present (%)	*OCD-like lesion absent (%)*	*p value*
Diagnosis [no. (%)]			
Early-onset Blount	4 (19)	17 (81)	
Late-onset Blount	3 (7)	39 (93)	0.21
Gender [no. (%)]			
Male	3 (7)	38 (93)	
Female	4 (18)	18 (82)	0.23
Mean age at X-rays (year)	8.1 (3.9–12.2)	11.1 (10.1–12.2)	0.06
(95%CI) (range)	(3–13.7)	(2–18.3)	
Blount disease [no. (%)]			
Right	2 (13)	14 (87)	0.07
Left	0 (0)	23 (100)	
Bilateral*	5 (21)	19 (79)	
Mean BMI (range)	21 (18–25)	36 (16–62)	0.003

*Two patients with bilateral Blount had OCD-like lesion noted on one extremity only

Table 4: Comparison of radiographic deformity parameters between patients with and without OCD-like lesions

	OCD-like lesion present mean (range)	*OCD-like lesion absent mean (range)*	*p value*
Plain X-rays			
MAD (mm)	63.3 (18.3–93.9)	71.9 (11.8–159.9)	0.39
mLDFA (°)	91.3 (87.0–100.0)	89.7 (80.5–108.0)	0.43
MPTA (°)	71.7 (48.0–83.9)	71.8 (42.0–87.0)	0.95

MAD, mechanical axis deviation; mLDFA, (mechanical) lateral distal femoral angle; MPTA, medial proximal tibial angle; OCD, osteochondritis dissecans



Figs 4A and B: Preoperative AP radiograph (A) in a 7-year-old girl with early-onset Blount disease demonstrates an OCD-like lesion involving the medial femoral condyle. Postoperative AP radiograph (B) taken 4 years after proximal lateral tibial hemiepiphysiodesis (using guided growth with extra-periosteal plate with subsequent hardware removal) demonstrates complete resolution of lesion

their aetiology, natural history and management remains unclear. We found no statistically significant difference in the prevalence of these lesions based on the child's age, gender, laterality, and magnitude of the varus deformity. None of the patients with the OCD-like lesion had any documented complaints of mechanical symptoms such as catching or locking, which may be suggestive of an unstable lesion, as was confirmed with intact overlying articular cartilage on MRI.[9] Based on the available information, none of these OCD-like lesions became symptomatic over time, required surgical intervention or increased in size.

The prevalence of OCD-like lesions in Blount disease at 10–19% based on different imaging studies is substantially higher than the 9.5/100,000 (0.0095%) reported for juvenile OCD.[10] The true aetiology of juvenile OCD of the knee remains unknown, although a mechanical trigger related to repetitive microtrauma to the knee is thought to play a critical role, with other potential causes being familial predisposition and local ischaemia to a vulnerable portion of the subchondral bone.[11] While both lesions are commonly found in the posterior aspect of the medial femoral condyle, the OCD-like lesions in our series involved primarily the central weight-bearing portion of the medial femoral condyle, as opposed to the slightly more lateral position, adjacent to the intercondylar notch in the classic OCD.[8] In a study by Chow et al., a narrower intercondylar notch width was implicated as a risk factor for juvenile OCD lesions.[12] They postulated that this anatomic risk factor might increase the likelihood for tibial eminence impingement and contribute to an OCD lesion. In a radiographic study of 103 knees using full-length standing radiographs, Jacobi et al. found an association between medial condyle OCD and varus malalignment of the knee.[13] However, we were unable to find any such relationship between the magnitude of varus deformity and presence of OCD-like lesions in our cohort of children with Blount disease.

Another possibility would be these OCD-like lesions represent a variation of the secondary ossification centres normally seen in the skeletally immature distal femur. Jans et al. found substantial

variability in the appearance of secondary ossification centres of the distal femur in 910 children and adolescents using MRI and noted that such variability was most often seen in the posterior third of the medial femoral condyle in young children.[14] Gebarski et al.[15] evaluated the MRIs of 38 children and looked for features that might distinguish between *in situ* OCD and normal variants of ossification at the distal femoral epiphysis based on Bohndorf's classification.[6] They concluded that location in the inferocentral posterior femoral condyles with intact overlying articular cartilage, accessory ossification centres, spiculations, residual cartilaginous model, and lack of bone marrow oedema were features more consistent with a normal variant, rather than a true OCD.

In a previous study, we had described the intra-articular MRI findings of children with Blount disease[3] including hypertrophy of the medial meniscus, especially the posterior horn, along with increased thickness of the unossified portion of the affected medial proximal tibial epiphysis. While not proven, the meniscal hypertrophy was thought to be compensatory to the skeletal deficiency secondary to the "sick" posteromedial proximal tibial physis which leads to the varus and procurvatum deformity of the proximal tibia. Perhaps, the juxtaposition of the OCD-like changes in the posterior aspect of the medial femoral condyle may also be related to the vulnerability of the underlying subchondral portion of the medial femoral condyle to the abnormal compressive forces. It is well known that excessive compressive forces can delay ossification of the epiphyseal cartilage, and this is possibly related to impaired vascularity to the growing epiphysis.[7] On the contrary, we did find lower BMI among children who had the OCD-like lesion compared with those without such findings. Additionally, two patients with bilateral Blount disease demonstrated OCD-like changes on one side only. Thus, while there are several plausible reasons for these OCD-like lesions in children with Blount disease, we were unable to confirm the true cause of these currently asymptomatic lesions.

There are several limitations in our study. First, given the retrospective nature of our study, only 37/63 (59%) of the patients with plain radiographs had an accompanying MRI. It is quite likely that had all 63 patients undergone an MRI, the prevalence of OCD-like lesions based on MRI may have been different. Given the added expense and possibility of requiring sedation, use of MRI in children with Blount disease is not routinely performed. Although we did not consistently get tunnel views of the knee, perhaps our yield for diagnosing OCD-like lesions on plain radiographs would have been even higher. Second, while we did have a relatively large cohort of patients, our study may be underpowered for detecting statistically significant associations between some of the demographic factors, including the magnitude of varus deformity. Additionally, none of the patients required specific treatment for the OCD-like lesions; routine follow-up imaging suggests that the natural history is for these lesions to resolve over time. Finally, given the intact articular cartilage overlying the OCD-like lesions of the medial femoral condyle, we did not perform any targeted surgical intervention for these lesions and thus did not have the opportunity to study their histopathology.

Conclusion

The OCD-like lesions of the ipsilateral medial femoral condyle are not uncommon in children with Blount disease. Based on the numbers available, we were unable to demonstrate any associations between the presence of this OCD-like lesion and the patient's age, gender, or magnitude of varus deformity. While these lesions have some features resembling previously described X-ray and MRI characteristics of irregular but normal variants of distal femoral epiphyseal ossification, there are some distinctions that more closely mimic true OCD lesions, including sagittal plane location (typically in the posterior aspect of the weight bearing portion of the medial femoral condyle) and associated bone marrow oedema. Normal variants of ossification affect the non-weight bearing portion of the condyle and are not associated with marrow oedema. Our data suggest that these lesions do not get symptomatic or enlarge over time. Furthermore, following realignment of the lower extremity, majority of these lesions do resolve over time. Further research is needed to fully ascertain the aetiology and natural history of these seemingly benign lesions in children with Blount disease.

References

1. Sabharwal S, Lee J Jr, Zhao C. Multiplanar deformity analysis of untreated Blount disease. Erratum appears in J Pediatr Orthop 2007;27(4):483 J Pediatr Orthop 2007;27(3):260–485. DOI: 10.1097/BPO.0b013e31803433c3.
2. Sabharwal S. Blount disease. J Bone Joint Surg Am 2009;91(7):1758–1776. DOI: 10.2106/JBJS.H.01348.
3. Sabharwal S, Wenokor C, Mehta A, et al. Intra-articular morphology of the knee joint in children with Blount disease: a case-control study using MRI. J Bone Joint Surg Am 2012;94(10):883–890. DOI: 10.2106/JBJS.K.00956.
4. Dipaola JD, Nelson DW, Colville MR. Characterizing osteochondral lesions by magnetic resonance imaging. Arthroscopy 1991;7(1):101–104. DOI: 10.1016/0749-8063(91)90087-e.
5. Hefti F, Beguiristain J, Krauspe R, et al. Osteochondritis dissecans: a multicenter study of the european pediatric orthopedic society. J Pediatr Orthop B [Clinical Trial Multicenter Study] 1999;8(4):231–245. DOI: 10.1097/01202412-199910000-00001.
6. Bohndorf K. Osteochondritis (osteochondrosis) dissecans: a review and new MRI classification. Eur Radiol [Review] 1998;8(1):103–112. DOI: 10.1007/s003300050348.
7. Trueta J, Trias A. The vascular contribution to osteogenesis. IV. The effect of pressure upon the epiphysial cartilage of the rabbit. J Bone Joint Surg Br 1961;43-B(4):800–813. DOI: 10.1302/0301-620X.43B4.800.
8. Caffey J, Madell SH, Royer C, et al. Ossification of the distal femoral epiphysis. J Bone Joint Surg Am 1958;40(3):647–654. DOI: 10.2106/00004623-195840030-00013.
9. Pascual-Garrido C, Moran CJ, Green DW, et al. Osteochondritis dissecans of the knee in children and adolescents. Curr Opin Pediatr [Review] 2013;25(1):46–51. DOI: 10.1097/MOP.0b013e32835adbf5.
10. Kessler JI, Nikizad H, Shea KG, et al. The demographics and epidemiology of osteochondritis dissecans of the knee in children and adolescents. Am J Sports Med 2014;42(2):320–326. DOI: 10.1177/0363546513510390.
11. Nawata K, Teshima R, Morio Y, et al. Anomalies of ossification in the posterolateral femoral condyle: assessment by MRI. Pedatr Radiol 1999;29(10):781–784. DOI: 10.1007/s002470050694.
12. Chow RM, Guzman MS, Dao Q. Intercondylar notch width as a risk factor for medial femoral condyle osteochondritis dissecans in skeletally immature patients. J Pediatr Orthop 2016;36(6):640–644. DOI: 10.1097/BPO.0000000000000511.
13. Jacobi M, Wahl P, Bouaicha S, et al. Association between mechanical axis of the leg and osteochondritis dissecans of the knee: radiographic study on 103 knees. Am J Sports Med 2010;38(7):1425–1428. DOI: 10.1177/0363546509359070.
14. Jans LB, Jaremko JL, Ditchfield M, et al. Evolution of femoral condylar ossification at MR imaging: frequency and patient age distribution. Radiology 2011;258(3):880–888. DOI: 10.1148/radiol.10101103.
15. Gebarski K, Hernandez RJ. Stage-I osteochondritis dissecans versus normal variants of ossification in the knee in children. Pediatr Radiol 2005;35(9):880–886. DOI: 10.1007/s00247-005-1507-6.