

Multiplanar Deformity Analysis of Untreated Blount Disease

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Abstract: Although varus malalignment of the proximal tibia is the primary pathology in Blount disease, other deformities may exist. To assess multiplanar lower limb deformities, children with previously untreated early- and late-onset Blount disease who subsequently needed surgical correction were identified. Preoperative frontal and sagittal plane deformity analysis using Paley's methodology and rotational profile assessment using prone clinical examination were performed by a single examiner. Results were compared between the 2 groups and with uninvolved limbs within each group. Additionally, rotational profile of the lower limb was compared with age-matched values. Over an 8-year period, 60 limbs (40 patients) including 26 with early-onset and 34 with late-onset Blount met the inclusion criteria. Although both groups exhibited proximal tibial varus, procurvatum, and internal torsion, patients with early-onset Blount disease had greater severity. Unlike the younger patients, approximately one third of the varus malalignment of the affected extremity was attributed to the distal femur in the late-onset patients. Neither group showed any significant deformity of the proximal femur and distal tibia or sagittal plane deformity of the distal femur. There was a correlation between the severity of varus malalignment of the limb with magnitude of proximal tibial deformities in both groups and with distal femoral varus in the late-onset group of patients. Multiplanar deformity analysis is a valuable tool in the comprehensive evaluation of children with Blount disease.

Key Words: deformity analysis, Blount disease, tibia vara, multiplanar deformity

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Natural history studies of Blount disease have suggested that progressive deformity can lead to gait abnormality, leg length inequality, and premature arthritis of the knee.^{1–3} Based on the Heuter-Volkman principle of increasing compressive forces causing growth inhibition of the posteromedial portion of the proximal tibial growth plate, a 3-dimensional deformity including varus, procurvatum, and internal rotation of the tibia, along with limb shortening can develop.^{4–6} Although an abnormality of the proximal tibia is the primary pathology in Blount disease, deformities in the

distal femur and distal tibia may be present in some patients.^{7–10} Unrecognized and thus uncorrected multiplanar abnormalities of the femur and tibia can lead to residual deformities, iatrogenic translational deformities,^{5,11} and increased incidence of “recurrence”, adding to the complexity of revision surgery.

Several authors have commented on the need for comprehensive treatment of the lower extremity in Blount disease.^{5,7,8} However, most studies have combined patients with and without previous surgeries and not outlined the differences in deformities, if any, based on age of disease onset. The goal of our study was to determine the location and magnitude of multiplanar deformities in patients with Blount disease who had not had any previous surgical or orthotic treatment. We also wanted to determine whether differences exist in the lower limb multiplanar deformities between patients with early- and late-onset forms of the disease.¹²

METHODS

After Institutional Review Board approval, the surgical database of the division of Pediatric Orthopaedics at our institution was searched for patients with the diagnosis of Blount disease from 1997 to 2005. Clinical charts and radiographic records were reviewed. Patients were excluded from the study if they had previous operative or orthotic treatment or did not have adequate clinical and radiographic records. The diagnosis of Blount disease was made using the following radiographic criteria: persistence or worsening genu varum with radiographic changes consistent with Blount disease¹³ and exclusion of any other etiology that would cause the observed physical or radiographic findings.

A retrospective review of outpatient records and radiographs was conducted. Demographic information including the patient's age when deformity was first noted was documented. Patients were classified as having early- or late-onset Blount disease based on onset of lower limb deformity before or after the age of 4 years¹² (Figs. 1 and 2). The rotational profile of both lower extremities measured on prone examination including hip rotation and thigh-foot angle¹⁴ at the final preoperative visit was noted. If the thigh-foot angle was internal, it was symbolized as negative and if external was marked as positive. Preoperative radiographic assessment included standing anteroposterior (AP) view of both lower extremities with the patellae pointing forward (Figs. 1B and 2B), AP and lateral views of the entire length of both tibias (Fig. 2C), and a scanogram. Deformity analysis was performed on the radiographs using methodology

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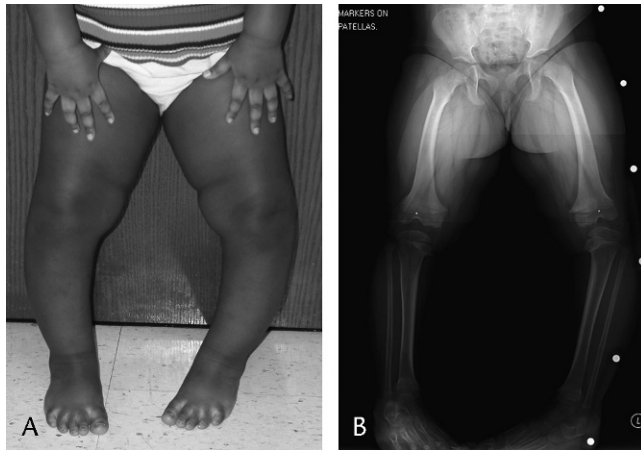


FIGURE 1. A, Clinical appearance of a 3-year-old girl with bilateral early-onset Blount disease, with the left side more severe than the right side with associated internal tibial torsion deformities. B, Standing AP radiograph of same patient with use of metal markers overlying the patellae to verify the knee forward position of the lower extremities. Radiographic magnification markers can be seen on the patients left side.

described by Paley et al.¹¹ Frontal plane analysis included mechanical axis deviation (MAD), lateral distal femoral angle mechanical (LDFA-m), medial proximal tibial angle (MPTA), neck-shaft angle (NSA), lateral distal tibial angle (LDTA), and joint line convergence angle (JLCA). Sagittal plane analysis included posterior distal femoral angle (PDFA), posterior proximal tibial angle (PPTA), and anterior distal tibial angle (ADTA). To improve measurement accuracy related to appropriate centering of the x-ray beam over the individual joints, NSA and LDTA were measured on

the hip (including the proximal third of the femoral shaft) and ankle (including the distal third of the tibial shaft) portions of the scanogram rather than the long-standing AP radiograph that was centered at the knee joint. Leg length discrepancy was measured in millimeters on the scanogram. The rotational profile and radiographic findings noted at the final preoperative visit were used for data analysis in this cross-sectional study.

Thirty-seven (93%) of the 40 patients, including 55 (92%) of the 60 affected limbs were clinically examined by a single fellowship-trained pediatric orthopaedist (S.S.). Any previously noted radiographic measurements were ignored, and all radiographs were remeasured for this study. All radiographic measurements were also performed by the same examiner (S.S.). Intraobserver reliability was calculated using a random set of 18 radiographs that were measured a few weeks apart by the same examiner.

Clinical rotational profile of the affected extremity was compared with the unaffected limb as well as age appropriate normal values.¹⁴ Because the “normal” population values of radiographic limb alignment parameters are based on a sample of adult subjects only,^{11,15} we were unable to use existing population controls for comparison of the radiographic alignment measures. The radiographic parameters of the unaffected limb of patients with unilateral disease were used as a control.

For statistical analyses, the *t* test was used to compare the frontal plane, sagittal plane, and rotational profile measurements between the control and affected limbs. The *t* test was also used to compare each of the deformity parameters in patients with early- versus late-onset forms of Blount disease. Correlation of MAD with LDFA-m, MPTA, PPTA, and thigh-foot angle was measured in both groups using the Pearson correlation coefficient (*r*). The level of

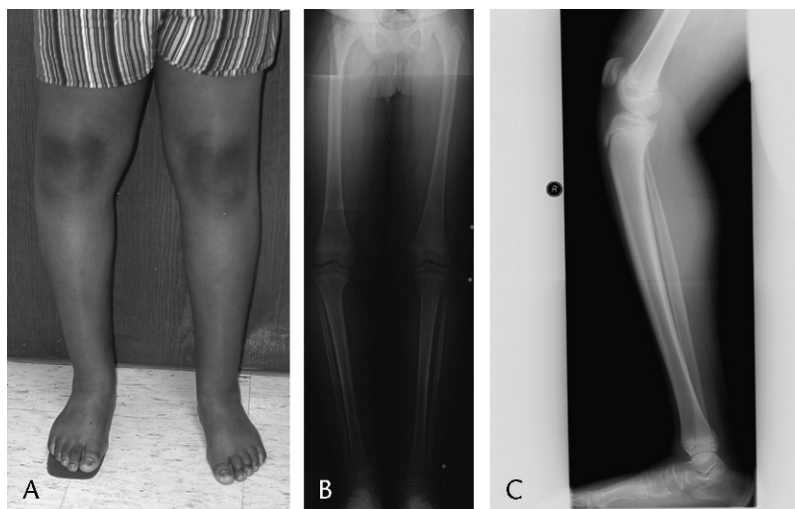


FIGURE 2. A, Clinical appearance of a 12.6-year-old boy with unilateral late-onset Blount disease. Note the lift under the right foot to compensate for shortening of the affected limb. B, Standing AP radiograph of the same patient. He had 60 mm of medial MAD on the right side. His LDFA-m measured 92 degrees on the right side and 86 degrees on the left side. Medial proximal tibial angle was 78 degrees on the right side and 88 degrees on the left side. C, Lateral view of the right tibia demonstrating 19 degrees of procurvatum deformity with a PPTA of 61 degrees.

significance was set at $P < 0.05$. All analyses were performed with SAS for Windows 9.1 software (SAS Institute Inc, Cary, NC).

RESULTS

Over the 8-year period, 40 patients with 60 affected limbs met the inclusion criteria for the current study. Demographic details are listed in Table 1. There were 16 children (26 limbs) in the early-onset group and 24 (34 limbs) in the late-onset group. Bilateral involvement was present in 62% of early-onset group and 42% of late-onset group. There was a predominance of girls in the early-onset group and boys in the late-onset group. The average age of onset of deformity was 2.3 years for the early-onset group and 10.8 years for late-onset group. Of the late-onset group of patients, 6 were classified as having juvenile and 18 as adolescent forms of Blount disease.¹² The average age at final preoperative visit was 4.5 years for the early-onset group and 12.7 years for the late-onset group.

The intraobserver reliability for measurement of all deformity parameters was good, with the intraclass correlation coefficients ranging from 0.87 to 0.89. Analysis of frontal plane deformities (Table 2) demonstrated an average medial MAD of 70 mm in the affected compared with 11 mm in the control limbs ($P = 0.003$) of the early-onset group and 71 mm compared with 2 mm in the control limbs ($P < 0.0001$) of the late-onset group. Average LDFA-m was 100 degrees compared with 101 degrees in the control limbs ($P = 0.89$) of the early-onset group and 94 degrees compared with 88 degrees in the control limbs ($P < 0.0001$) of the late-onset group. Average MPTA was 68 degrees compared with 91 degrees in the control limbs ($P = 0.005$) of the early-onset group and 75 degrees compared with 87 degrees in the control limbs ($P < 0.0001$) of the late-onset group. The average JLCA was 15 degrees medial compared with 3 degrees medial in the control limbs ($P = 0.03$) of the early-onset group and 2 degrees medial compared with 1 degree medial in the control limbs ($P = 0.10$) of the late-onset group. The average NSA and LDTA was similar to the control limbs for both early- and late-onset groups. Radiographic measurement of limb length discrepancy in the unilateral cases revealed an average shortening in the affected limb of 17 mm in the early-onset group and 19 mm in the late-onset group.

Analysis of sagittal plane deformities (Table 3) demonstrated no significant difference in distal femoral

TABLE 1. Patient Demographics

	Early Onset	Late Onset	Total
No. patients	16	24	40
No. limbs	26	34	60
Unilateral/bilateral	6:10	14:10	20:20
Male/female	3:13	19:5	22:18
Age at onset (y), mean (range)	2.3 (1.1–3.5)	10.8 (6–14)	7.3 (1.1–14)
Age at examination (y), mean (range)	4.5 (1.8–8.2)	12.7 (8.7–15.9)	9.4 (1.8–15.9)

TABLE 2. Radiographic Measurement of Frontal Plane Deformities

	Early Onset		Late Onset	
	Group 1: Control	Group 2: Blount	Group 3: Control	Group 4: Blount
MAD (mm)	11 (–5–17)	70* (55–85)	2 (–6–10)	71† (60–82)
LDFA-m (degrees)	101 (88–114)	100 (95–104)	88 (87–89)	94‡ (93–95)
MPTA (degrees)	91 (87–95)	68* (62–74)	87 (86–89)	75‡ (73–78)
JLCA (degrees)	3 (–5–11)	15* (11–20)	1 (0–2)	2‡ (1–4)
NSA (degrees)	146 (133–159)	150 (145–155)	143 (139–147)	143‡ (140–146)
LDTA (degrees)	90 (88–92)	91 (88–95)	87 (84–89)	85‡ (84–87)

Data are presented as mean (95% confidential interval).
MAD: medial (+); lateral (–).
JLCA: medial (+); lateral (–).
*There was significant difference between groups 1 and 2, $P < 0.05$.
†There was significant difference between groups 3 and 4, $P < 0.05$.
‡There was significant difference between groups 2 and 4, $P < 0.05$.

alignment (PDFA) of the affected compared with the control limbs for both early- and late-onset groups. However, there was significant procurvatum deformity of the proximal tibia, indicated by lower PPTA compared with control limbs in both groups. The average PPTA was 64 degrees compared with 82 degrees in the control limbs ($P = 0.002$) of the early-onset group and 71 degrees compared with 78 degrees in the control limbs ($P = 0.002$) of the late-onset group. The average ADTA was similar to control limbs in both groups.

Analysis of rotational profile is shown in Table 4. Hip rotation of the affected extremity was similar to that of the unaffected limb and age-appropriate population values¹⁴ for the early-onset group with slight asymmetry in the late-onset group. Differences in thigh-foot angle were more evident in the early-onset group, with greater internal tibial torsion of the affected extremity.

Correlation of MAD with various deformity parameters is shown in Table 5. There was a strong correlation between varus malalignment of the limb (medial MAD) and

TABLE 3. Radiographic Measurement of Sagittal Plane Deformities

	Early Onset		Late Onset	
	Group 1: Control	Group 2: Blount	Group 3: Control	Group 4: Blount
PDFA (degrees)	84 (74–94)	83 (81–85)	86 (84–88)	85 (82–88)
PPTA (degrees)	82 (81–83)	64* (55–73)	78 (75–81)	71† (68–73)
ADTA (degrees)	91 (86–96)	90 (88–91)	87 (84–90)	87‡ (85–89)

Data are presented as mean (95% confidential interval).
*There was significant difference between groups 1 and 2, $P < 0.05$.
†There was significant difference between groups 3 and 4, $P < 0.05$.
‡There was significant difference between groups 2 and 4, $P < 0.05$.

TABLE 4. Analysis of Rotational Profile Based on Prone Clinical Examination

	Early Onset			Late Onset		
	Group 1: Normal*	Group 2: Control	Group 3: Blount	Group 4: Normal*	Group 5: Control	Group 6: Blount
Hip internal rotation (degrees)	47 (43 to 51)	43 (8 to 78)	58 (47 to 68)	45 (41 to 49)	27† (16 to 37)	34‡§ (25 to 44)
Hip external rotation (degrees)	52 (49 to 57)	53 (29 to 77)	46 (38 to 55)	40 (36 to 44)	54† (44 to 63)	58‡§ (52 to 63)
Thigh-foot angle (degrees)	8 (6 to 10)	4 (1 to 7)	-23 ¶ (-30 to -16)	19 (13 to 25)	10† (3 to 16)	-3‡§# (-8 to 1)

Data are presented as mean (95% confidential interval).
 Thigh-foot angle: external (+); internal (-).
 *Normal values based on references 14.
 †There was significant difference between groups 1 and 3, $P < 0.05$.
 ‡There was significant difference between groups 2 and 3, $P < 0.05$.
 §There was significant difference between groups 3 and 6, $P < 0.05$.
 ||There was significant difference between groups 4 and 5, $P < 0.05$.
 ¶There was significant difference between groups 4 and 6, $P < 0.05$.
 #There was significant difference between groups 5 and 6, $P < 0.05$.

increasing varus deformity of the tibia (lower MPTA) for both the early- and late-onset groups of patients. A strong correlation of MAD was also noted with varus deformity of the distal femur (higher LDFA-m) for the late-onset but not the early-onset group. Greater sagittal plane deformity of the tibia, that is, procurvatum (lower PPTA) was seen with increasing varus malalignment of the limb in early-onset and not the late-onset group of patients.

DISCUSSION

The etiology of Blount disease remains controversial, although most agree that it is related to abnormal compressive forces across the proximal tibial growth plate, causing local growth inhibition, and secondary deformities.^{12,16-18} Wenger et al¹⁸ suggested that the proximal tibial growth plate responds differently at different ages, with increased pliability in the unossified epiphyses seen in the younger patients causing more growth inhibition compared with adolescents. This is consistent with our observation of increased magnitude of deformities in all 3 planes in the younger patients. Some authors have advocated use of arthrography^{19,20} and magnetic resonance imaging^{20,21} to delineate the proximal tibial deformity, especially in early-onset disease, because plain radiographs may overestimate the “depression” of the medial plateau. This may be related to delayed ossification of the affected medial tibial epiphyses secondary to increased compressive forces²² and perhaps contributed to the greater proximal tibial deformity observed in the early-onset group.

The goal of treatment in Blount disease is to obtain a well-aligned lower extremity with normal joint orientation and equal leg lengths that are maintained beyond skeletal maturity. To avoid undercorrection, overcorrection, and creation of iatrogenic deformities, evaluation of limb deformities at locations besides the proximal tibia is vital in patients with Blount disease. There is no consensus in the literature regarding the ideal alignment of the lower extremity after surgical reconstruction for Blount disease. Some advocate normalization of the mechanical axis,²³ whereas others feel that some degree of overcorrection should be attempted.²⁴⁻²⁶ A significant drawback in many such reports is that the assessment of postsurgical limb alignment is based on limited non-weight-bearing radiographs of the knee, without visualization of the entire length of the femur and tibia. Especially when compounded with an overlying cast, detailed analysis of MAD and joint orientation parameters may not be feasible. Overcorrection with a valgus osteotomy at the proximal tibia can also create secondary deformities at the distal tibia and produce malorientation of the ankle joint.^{5,11}

Despite being referred to as tibia vara,²⁷ Blount disease may have other sources of medial MAD including the distal femur^{5,8-10} and intraarticular deformities, creating a dynamic varus thrust.⁶ If there is a substantial varus deformity of the distal femur in a patient with Blount disease and the medial MAD (ie, varus malalignment) is fully corrected with a single proximal tibial osteotomy, malorientation of the knee joint can result. The resultant knee joint obliquity can generate abnormal translational and shear forces and may lead to premature osteoarthritis of the knee. To avoid significant knee joint obliquity, Gordon et al⁸ used an LDFA-m greater than 95 degrees in patients with late-onset Blount disease as an indication for correcting the distal femoral varus deformity. However, we are not aware of any long-term studies that have correlated varying degrees of residual knee joint obliquity with generation of shear forces or development of degenerative arthritis in such patients. Chao et al,¹⁵ reporting on healthy adults, noted less than 2 degrees of knee joint obliquity on standing radiographs. Cooke²⁸ suggested an association between knee joint obliquity and osteoarthritis in

TABLE 5. Correlation Between MAD and Other Deformity Variables

	Early Onset		Late Onset	
	r	P	r	P
LDFA-m	0.02	0.89	0.53	0.002
MPTA	-0.70	<0.0001	-0.86	<0.0001
PPTA	-0.83	0.0005	-0.18	0.44
Thigh-foot angle	-0.32	0.13	-0.45	0.01

r is correlation coefficient. P is p value.

the adult population. Although making empirical sense, the issue of limits of tolerance of knee joint obliquity remains unresolved.

The few long-term studies on the natural history and treatment of Blount disease¹⁻³ are limited by the lack of appropriate biplanar radiographs necessary for analysis of sagittal plane deformities, as well as absence of detailed evaluation of joint orientation and rotational deformities of the lower limb. Hofmann et al,¹ reporting on patients with early-onset Blount disease who had undergone realignment surgery 12 years earlier, found that more than half of the knees had premature degenerative arthritis. Interestingly, 7 of the 11 symptomatic knees had “five to ten degrees of flexion contracture.” Most of these symptomatic individuals underwent knee arthroscopy and degenerative arthritis of the lateral compartment along with posterior depression of the medial tibial plateau was observed. Although there was no mention of lateral radiographs in this report, the flexion “contracture” was likely bony and not soft tissue in origin. The flexion deformity may be secondary to an unrecognized and thus untreated procurvatum of the proximal tibia. These persistent deformities may have lead to asymmetric loads across the knee joint and likely contributed to the development of premature arthritis.

Based on our review of the literature, this is the first report with in-depth analyses of multiplanar deformities in patients with either early- or late-onset Blount disease who have not had previous treatment. However, there are a few limitations in our study. First, given the lack of complete ossification of the proximal tibial epiphyses, especially in the early-onset group, the magnitude of proximal tibial deformity may have been overestimated on the plain radiographs. Further evaluation of the proximal tibial deformity with imaging modalities such as arthrography or magnetic resonance imaging would be useful before performing surgical realignment of the extremity. Second, consistent with previous reports,^{7,8} we found a procurvatum deformity of the proximal tibia in Blount disease. However, correlation of this radiographic finding with clinically apparent flexion deformity of the knee was not reported because of the retrospective nature of our study. Third, analysis of limb rotation was done on clinical examination rather than with computed tomography and may be subject to a greater degree of measurement error, especially in obese patients with associated varus malalignment of the knee. Fourth, although this may be one of the larger studies reporting on Blount disease, given the multiple deformity variables that were analyzed, there is a possibility of a type II error²⁹ when interpreting results that did not achieve statistical significance. Finally, although our findings support the practice of systematic multiplanar analysis of deformities in Blount disease, surgical outcome after realignment was not the subject of this study. Thus, based on our results, we are unable to make specific recommendations on surgical treatment.

In summary, although both groups of patients in our study exhibited multiplanar deformities of the proximal tibia including varus, procurvatum, and internal torsion, children with early-onset Blount disease had greater severity compared with the late-onset group. In children with late-onset disease,

approximately one third of the varus malalignment of the limb was attributed to the distal femur. There was a strong correlation of varus malalignment of the limb with increasing proximal tibial deformity in both groups as well as with increasing varus deformity of the distal femur in the late-onset group of patients. Neither group showed any significant frontal plane deformity of the proximal femur and distal tibia or sagittal plane deformity of the distal femur and distal tibia. Limb shortening of similar magnitude was noted in both groups of patients. Thus, in order to individualize treatment and avoid iatrogenic deformities, a comprehensive multiplanar deformity analysis should be routinely performed when evaluating a child with Blount disease.

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