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# A comparative evaluation of tibial metaphyseal-diaphyseal angle changes between physiologic bowing and Blount disease

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**Abstract**

The purpose of this study was to estimate the rate of spontaneous improvement in tibial metaphyseal-diaphyseal angle (TMDA) in physiologic bowing in comparison to that in Blount disease and to provide reference values of TMDA for monitoring patients with highly suspected to have Blount disease.

We retrospectively reviewed patients with physiologic bowing meeting the following criteria:
(1) TMDA greater than 9° before 36 months of age at initial evaluation;
(2) two or more standing long bone radiographs available; and
(3) follow-up conducted up to resolution of deformity.

Patients with Blount disease had
(1) more than 2 standing long bone radiographs obtained before 36 months of age and
(2) underwent no treatment during the period in which these images were obtained.

TMDA measurements were obtained from 174 patients with physiologic bowing and 32 patients with Blount disease. Rates of TMDA improvement were adjusted by multiple factors using a linear mixed model, with sex and laterality as fixed effects and age and individual patients as the random effects.

In the physiologic bowing group, TMDA improved significantly, by 3° per 6 months and by 6° per year. Changes in TMDA were not significant in the Blount disease group.

Knowing the rate of TMDA change can be helpful for physicians seeking to monitor infants with suspected as having Blount disease with a high TMDA and to avoid unnecessary repeat radiographic evaluations.

**Abbreviation:** TMDA = tibial metaphyseal diaphyseal angle.

**Keywords:** blount disease, physiologic bowing, tibia vara, tibial metaphyseal diaphyseal angle

## 1. Introduction

Distinguishing between physiologic bowing and Blount disease is very difficult in children between 1 and 3 years of age based on only one radiograph at the initial visit.[1–3] Early and accurate distinction between these 2 conditions is of great importance because while physiologic bowing does not require treatment, Blount disease should be treated with bracing as soon as possible.[4,5] Radiographic changes on the medial side of the proximal tibial physis characteristic of Blount disease were initially described by Langenskiold, and in cases where these changes are observed, the diagnosis is easily established.[1,6] However, if no changes are seen at the growth plate, it can be impossible to distinguish physiologic bowing from Blount disease using a single radiograph.

A few radiologic parameters have been developed to address this diagnostic challenge. Although the tibial metaphyseal-diaphyseal angle (TMDA) has classically been used to differentiate between physiologic bowing and Blount disease,[2,7] several studies have questioned the predictive value of the TMDA.[7–9] Some studies have reported that the ratio of the femoral to tibial metaphyseal-diaphyseal angles may be a more accurate predictor than the TMDA,[3] although there are no reports on the clinical application or usefulness of this ratio. Thus, ultimately, follow-up radiographs may be necessary in patients with suspected as having Blount disease with a high TMDA until the physician can rule out Blount disease. Additionally, even if physicians observe the improvement in TMDA on follow-up radiograph, it is still difficult to confirm physiologic bowing.

Therefore, the purpose of this study was to estimate the rate of spontaneous improvement in TMDA in physiologic bowing in comparison to that in Blount disease through the application of a linear mixed model and to provide reference values of TMDA for monitoring patients with suspected as having Blount disease. We

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hypothesized that quantifying the degree of improvement or progression of the TMDA with age might be helpful for physicians trying to distinguish between Blount disease and physiologic bowing during follow up.

## 2. Materials and methods

This retrospective study was approved by the Institutional Review Board of our hospital (IRB No. 4-2016-0668). An electronic search of medical records from the period between March 2003 and December 2014 identified a total of 975 consecutive patients who had been evaluated for bowed-leg deformity prior to an age of 48 months. To reduce the risk of false positive error when distinguishing between physiologic bowing and Blount disease, we used a TMDA greater than 9° as a dividing line based on published criteria.[7] At our institution, all patients with a TMDA greater than 9° had been followed at 3 to 6-month intervals until bowing resolved. If the TMDA showed improvement with age, the children were diagnosed as having physiologic bowing. The inclusion criteria for the physiologic bowing group were as follows:

1. TMDA greater than 9° before the age of 36 months at initial evaluation,
2. availability of more than 2 standing long bone radiographs of the affected lower extremity before the age of 36 months, and
3. minimum of 1 year of follow up with documented radiologic resolution of deformity confirming a final diagnosis of physiologic bowing at last follow-up.

We excluded any patient with a bowed-leg deformity of neuromuscular, congenital, metabolic, or traumatic etiology.

We then identified patients with Blount disease to compare changes in TMDA in this group vs those in the physiologic bowing group. For the same time period, thirty-two infants diagnosed with Blount disease were identified. All patients were treated with brace treatment or surgical treatment. At our institution, brace treatment is applied in patients showing radiographic signs such as lucency, sclerosis, or fragmentation of the medial portion of the proximal tibia metaphysis or an increase in TMDA during follow up. The inclusion criteria for the Blount disease group were as follows:

1. confirmed with Blount disease regardless of the initial TMDA
2. younger than 36 months old when first evaluated at our institution;
3. availability of more than 2 standing long bone radiographs of the affected lower extremity before the age of 36 months without any treatment.

We excluded patients who had received brace treatment at other hospitals.

For all patients included in the study, initial and follow-up radiographs comprised standing long bone anteroposterior views of the lower extremity from the hip to the ankle, with the patella forward. As described by Levine and Drennan, the TMDA was measured on all radiographs as the angle between a line perpendicular to the lateral border of the tibial cortex and a line connecting the lateral and medial beaks of the proximal tibial metaphysis (Fig. 1).[2]

All radiologic measurements were performed by 2 orthopedic clinical fellows who were blinded to the study. All measurements were made independently, twice for each radiograph, and with at least 6 months between assessments.


**Figure 1.** Representation of the tibial metaphyseal-diaphyseal angle (TMDA). A line drawn perpendicular to the lateral border of the tibial cortex and another is drawn through the 2 beaks of the metaphysis to determine the transverse axis of the tibial metaphysis. The TMDA is the angle bisected by those 2 lines.

## 3. Statistical analysis

In this study, a linear mixed model was applied to estimate patterns of spontaneous improvement in TMDA over time in children with physiologic bowing. The model assumed normally distributed errors and included both fixed and random effects, such as analysis of variance (ANOVA) incorporating a random effect.[10] The main application of this model is usually to characterize ways in which an outcome changes over time and predictors of such change;[11] it is particularly useful in longitudinal studies designed to investigate changes over time where a given characteristic is measured repeatedly for each patient.[12,13] A linear mixed model was thus suitable for analyzing the serial data collected in this investigation: radiographic measurements obtained at multiple follow-up evaluations for children with bowed-leg deformities.

The rate of improvement in the TMDA was adjusted by multiple factors using a linear mixed model, with sex and laterality[14] as fixed effects and age and individual patients as random effects. The covariance structure was assumed as unstructured components. Restricted maximum likelihood estimation was used as the estimation method to produce unbiased estimators. A linear mixed model was built to estimate the rate of improvement by incorporating the linear age effect, sex, and laterality as covariates. In this model, the slope reflected the rate of improvement per month. Following examination of the individual pattern of the rate of angular correction along with the duration of follow up, a model with a random slope and a random intercept was suggested. Linear age, sex, and laterality effects were integrated to produce the estimation of the spontaneous improvement rate of the TMDA in patients with physiologic bowing. Statistical analyses were performed using SAS software (version 9.2, SAS Inc., Cary, NC). All statistics were 2-tailed, and $P$ values $< .05$ were considered significant. Inter- and intra-observer reliabilities were gauged, as well, using intraclass correlation coefficients (ICCs). ICCs were interpreted as follows: poor, less than 0.4; marginal, between 0.4 and 0.75; and good, greater than 0.75.

## 4. Results

A total of 174 patients (257 extremities) met the criteria for inclusion in the physiologic bowing group. There were 82 boys and 92 girls, and all patients were of Asian descent. In this group, both lower extremities were affected in 83 patients, while 91 patients had unilateral bowing. The mean patient age at initial radiographic evaluation was 17.3 (12–30) months. The mean follow-up duration was 18.4 (12–33) months. The mean interval between follow-up evaluations was 6.5 (3–22) months. The mean number of follow-up evaluations was 2.9 (2–7) visits. Measurements were obtained from a total of 748 radiographs.

The Blount disease comparison group comprised 32 patients (48 extremities). There were 21 boys and 11 girls. Bilateral limb deformity affected 16 patients. In the Blount disease group, the mean patient age at initial radiographic evaluation was 18.5 (12–29) months. The mean follow-up duration was 4.5 (4–8) years. The mean interval between follow-up evaluations was 4.6 (2–14) months. The mean number of follow-up evaluations was 2.4 (2–5) visits. A total of 107 radiographs were reviewed from patients in this group.

In the physiologic bowing group, the TMDA was found to improve significantly as patients grew older (Fig. 2). Among patients with physiologic bowing, the TMDA decreased by 0.496° (95% confidence interval (CI): 0.467°–0.526°, $P < .001$) per month (Table 1 and Fig. 3). Other covariates (e.g., sex and laterality) did not affect spontaneous improvement of the TMDA. In contrast, among patients in the Blount disease group, TMDA changes with age were not significant (Table 1 and Fig. 3). In the Blount disease group, the mean TMDA at initial visit was 13.9° (7.6°–20.4°), and 3 patients with TMDA less than 9° at initial evaluation were diagnosed as Blount disease on follow-up evaluation. In the physiologic bowing group, the TMDA was found to improve by 3° per 6 months and by 6° per year (Table 2).

Inter-rater reliability of radiographic TMDA measurements was confirmed using ICCs (Table 3). ICC ranges for intra-observer (0.901–0.944) and inter-observer (0.925–0.952) reliabilities were comparable to those in previous studies.[15,16] The mean absolute differences between duplicate measurements of TMDA by each observer were 2.2° and 1.7°. The mean absolute differences in measurements between 2 observers were 1.5° and 1.9°.

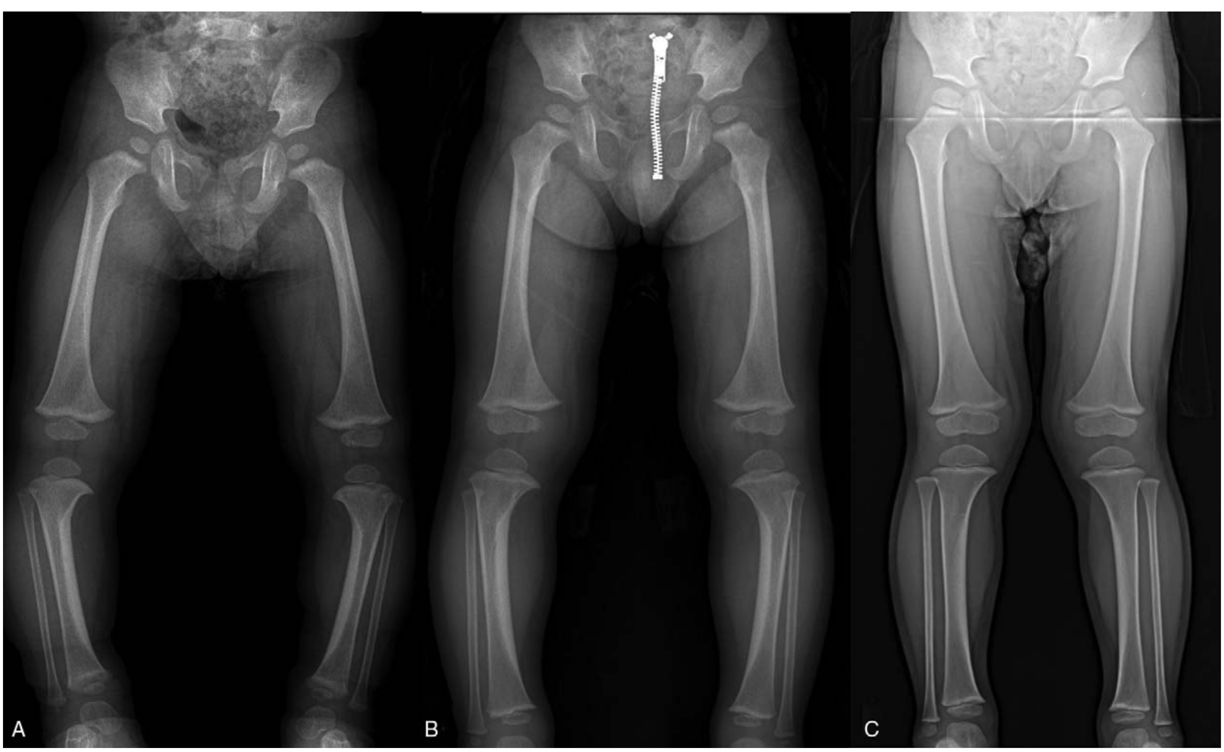


**Figure 2.** A 15-month-old boy presented with bilateral bow-leg deformities. (A) At the initial visit, radiographs revealed tibial metaphyseal-diaphyseal angles (TMDAs) of 13° on the right and 12° on the left. (B) There was slight improvement after 4 months, with TMDAs of 9° on the right and 10° on the left. (C) Radiograph showing definite improvement at the age of 31 months. The TMDA measured 0.5° on the right and 0.5° on the left.

**Table 1**

**Estimation of TMDA with use of linear mixed model including fixed and random effects.**

| | Physiologic bowing group | | | Blount disease group | | |
|---|---|---|---|---|---|---|
| | **Estimation (95% CI) (degrees)** | **SE** | ***P* value** | **Estimation (95% CI) (degrees)** | **SE** | ***P* value** |
| Intercept | 18.690 (16.413 to 20.966) | 1.153 | <.001 | 15.277 (9.088 to 21.467) | 3.026 | <.001 |
| Age | −0.496 (−0.526 to −0.467) | 0.015 | <.001 | −0.068 (−0.213 to 0.076) | 0.070 | .341 |
| Sex | −0.290 (−0.905 to 0.326) | 0.313 | .355 | 0.341 (−1.637 to 2.318) | 0.983 | .741 |
| Laterality | 0.386 (−0.625 to 1.396) | 0.514 | .453 | −0.352 (−2.353 to 1.650) | 0.995 | .725 |

CI = confidence interval, SE = standard error, TMDA = tibial metaphyseal diaphyseal angle.

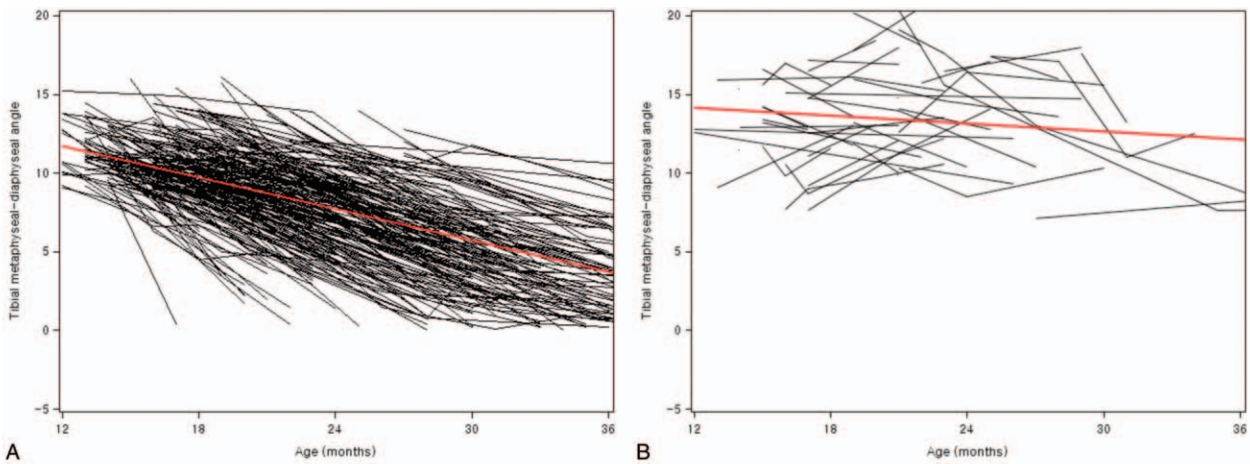


Figure 3. The reference values for tibial metaphyseal-diaphyseal angles (TMDAs) are shown in each group. The red lines represent estimates of improvement in the TMDA according to a linear mixed model. (A) Physiologic bowing group, (B) Blount disease group.

## 5. Discussion

Few studies have investigated the natural history of this condition.[17] We found that there were differences between the study groups in the rates of change in TMDA. In the physiologic bowing group, the TMDA decreased by 3° per 6 months and by 6° per year, and the only factor significantly contributing to these improvement rates was age. In contrast, in the Blount disease group, no improvement in TMDA was observed with age.

In the physiologic bowing group, TMDA decreased by a mean of 0.5° per month. Another previous study reported that TMDA decreased an average of 0.35° per month during the first year of observation in patients with physiological bowing,[7] which is a smaller value than our result. This discrepancy may reflect differences in study inclusion criteria for patients with physiologic bowing. While the previous study included patients with a TMDA less than 9°, we excluded these patients because most physicians do not follow them with serial radiographs due to the low possibility of Blount disease associated with such small angular deformities. Another reason for this difference may be the use of different statistical methods for analyzing changes in TMDA. In this

**Table 2**

**Estimation of TMDA by age (in months) from linear mixed model.**

| | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|
| Age (months) | 12 | 15 | 18 | 21 | 24 | 27 | 30 | 33 | 36 |
| TMDA (degrees) | 12.7 | 11.2 | 9.8 | 8.3 | 6.8 | 5.3 | 3.8 | 2.3 | 0.8 |
| 95% CI (degrees) | 10.5 to 14.9 | 9.1 to 13.4 | 7.8 to 11.9 | 6.1 to 10.4 | 4.6 to 9.0 | 3.1 to 7.5 | 1.6 to 6.0 | 0.1 to 4.5 | −1.4 to 3.0 |

CI = confidence interval, TMDA = tibial metaphyseal diaphyseal angle.

**Table 3**

**Reliability measurements of TMDA.**

| | Intraobserver | | | Interobserver | |
|---|---|---|---|---|---|
| **Observer** | **Mean difference (±SD)** | **ICC (95% CI)** | **Fellow A vs B** | **Mean difference (±SD)** | **ICC (95% CI)** |
| Fellow A | 2.2° ± 2.1° | 0.901 (0.836–0.943) | 1st measurement | 1.5° ± 1.7° | 0.952 (0.909–0.975) |
| Fellow B | 1.7° ± 1.4° | 0.944 (0.897–0.970) | 2nd measurement | 1.9° ± 1.9° | 0.925 (0.875–0.958) |

CI = confidence interval, ICC = intraclass correlation coefficient, SD = standard deviation, TMDA = tibial metaphyseal diaphyseal angle.

study, we used a linear mixed model to achieve a more reliable result.

Among patients in the Blount disease group, no significant change in TMDA was observed with age progression, and no other factors were found to have a significant influence on TMDA change. There is some controversy surrounding the progression of TMDA with age in Blount disease. Several studies have shown conflicting and inconsistent results with regards the changes in TMDA in Blount disease.[7,9,17,18] In this study, no significant change in TMDA with age indicated by the linear mixed model in Blount disease group.

Two techniques have been described for measurement of TMDA. These techniques involve using either the lateral border of the tibial cortex, as described by Levine and Drennan,[2] or the center of the tibial shaft as the longitudinal axis for radiologic measurements.[19] Previous studies have reported no statistical differences between measurements made using either of these 2 methods, and measurements of the TMDA using the method of Levine and Drennan have been reported to have better reproducibility.[15,19] Therefore, between the 2 techniques, we chose the method of Levine and Drennan. Although TMDA measurements have been proven to be reliable, standard deviations for measurements of the TMDA have been found to range from 2° to 2.8°.[15,16] In particular, tibial rotation has been reported to have potentially significant effects on the measured TMDA.[3,20] In our study, the mean differences between duplicate measurements for each observer were 2.2° and 1.7°, and the mean differences in measurements between 2 observers were 1.5° and 1.9°. These results were almost consistent with previous studies.[16] These issues should be considered by physicians when evaluating changes in TMDA during follow up.

Measurement of TMDA is not the only method used to diagnose physiologic bowing. There are clinical ways of measuring the intercondylar distance to avoid radiation exposure during follow up.[21] Some physicians observe patients with a TMDA lower than 10° to 12° without radiologic follow up until they are around 3 years of age. However, physicians may miss the proper timing of brace treatment for Blount disease using physical examination without radiographic evaluation. EOS imaging system can be a good option for patients with suspected Blount disease. EOS provides high-quality images of lower extremity and delivers low radiation dose that is 2 to 3 times less than a conventional X-ray.[22,23] Although EOS system is expensive and has several limitations,[24] it is a valuable tool for diagnosis and follow-up in pediatric orthopeadic area.

Notwithstanding, confirming the diagnosis of Blount disease without visualization of Langenskiold changes during the follow-up period is difficult. In addition, there are no specific guidelines for how these patients should be followed up, potentially subjecting them to unnecessary repeated radiographic evaluations. Considering the noted improvement in the TMDA of 3° per 6 months and the associated variability of measuring TMDA, we assume that patients highly suspected of having Blount disease should be followed at intervals longer than 9 months to obtain a more precise radiographic assessment. Additionally, the cut-off value of the TMDA for evaluation of bowed-leg deformity should be changed according to a patient's age at the time of presentation, because TMDA decreases with age.

Some limitations of this study should be noted. First, the number of patients with Blount disease was relatively small, compared to the number of patients with physiologic bowing. This was not unexpected because of the very low prevalence of Blount disease (less than 1% in infants and toddlers) and because we included only patients with Blount disease before brace treatment in our analysis. Nevertheless, the relatively small number of patients in the Blount disease group may have reduced the power of our statistical analysis. Second, body mass index and age at independent walking were not considered in our analysis as factors that might potentially impact spontaneous TMDA correction. Although these features have been identified as possible risk factors for Blount disease,[25–27] we could not include these factors in our model in this study. However, we believe that this limitation does not jeopardize our results because we focused only on the rate of spontaneous TMDA improvement. Third, it is important to note that our results should not be used as sole criteria for distinguishing between physiologic bowing and Blount disease, establishing diagnostic criteria using TMDA change with age requires further clinical investigations. Nonetheless, our results can be applied as an additional tool for interpreting radiographic measurements obtained at follow-up evaluations.

## 6. Conclusion

In patients with physiologic bowing, TMDA improved by 3° per 6 months as patients grew. No significant change in the TMDA over time was observed in the Blount disease group. Using the reference values of the TMDA, physicians can monitor infants highly suspected as having Blount disease with high TMDA and avoid unnecessary repeat radiographic evaluations.

## Author contributions

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## References

[1] Langenskiöld A, Riska EB. Tibia vara (osteochondrosis deformans tibiae). J Bone Joint Surg Am 1964;46:1405–20.
[2] Levine AM, Drennan JC. Physiological bowing and tibia vara. The metaphyseal-diaphyseal angle in the measurement of bowleg deformities. J Bone Joint Surg Am 1982;64:1158–63.
[3] McCarthy JJ, Betz RR, Kim A, et al. Early radiographic differentiation of infantile tibia vara from physiologic bowing using the femoral-tibial ratio. J Pediatr Orthop 2001;21:545–8.
[4] Sabharwal S. Blount disease: an update. Orthop Clin North Am 2015;46:37–47.
[5] Birch JG. Blount disease. J Am Acad Orthop Surg 2013;21:408–18.
[6] Khanfour AA. Does Langenskiold staging have a good prognostic value in late onset tibia vara? J Orthop Surg Res 2012;7:23.
[7] Feldman MD, Schoenecker PL. Use of the metaphyseal-diaphyseal angle in the evaluation of bowed legs. J Bone Joint Surg Am 1993;75:1602–9.
[8] Eggert P, Viemann M. Physiological bowlegs or infantile Blount's disease. Some new aspects on an old problem. Pediatr Radiol 1996;26:349–52.
[9] Hagglund G, Ingvarsson T, Ramgren B, et al. Metaphyseal-diaphyseal angle in Blount's disease. A 30-year follow-up of 13 unoperated children. Acta Orthop Scand 1997;68:167–9.

[10] Bolker BM, Brooks ME, Clark CJ, et al. Generalized linear mixed models: a practical guide for ecology and evolution. Trends Ecol Evol 2009;24:127–35.
[11] Cnaan A, Laird NM, Slasor P. Using the general linear mixed model to analyse unbalanced repeated measures and longitudinal data. Stat Med 1997;16:2349–80.
[12] Laird NM, Ware JH. Random-effects models for longitudinal data. Biometrics 1982;38:963–74.
[13] Nguyen DV, Senturk D, Carroll RJ. Covariate-adjusted linear mixed effects model with an application to longitudinal data. J Nonparametr Stat 2008;20:459–81.
[14] Park MS, Kim SJ, Chung CY, et al. Statistical consideration for bilateral cases in orthopaedic research. J Bone Joint Surg Am 2010;92:1732–7.
[15] Lavelle WF, Shovlin J, Drvaric DM. Reliability of the metaphyseal-diaphyseal angle in tibia vara as measured on digital images by pediatric orthopaedic surgeons. J Pediatr Orthop 2008;28:695–8.
[16] Henderson RC, Lechner CT, DeMasi RA, et al. Variability in radiographic measurement of bowleg deformity in children. J Pediatr Orthop 1990;10:491–4.
[17] Shinohara Y, Kamegaya M, Kuniyoshi K, et al. Natural history of infantile tibia vara. J Bone Joint Surg Br 2002;84:263–8.
[18] Johnston CE 2nd. Infantile tibia vara. Clin Orthop Relat Res 1990;255:13–23.
[19] Auerbach JD, Radomisli TE, Simoncini J, et al. Variability of the metaphyseal-diaphyseal angle in tibia vara: a comparison of two methods. J Pediatr Orthop 2004;24:75–8.
[20] Stricker SJ, Faustgen JP. Radiographic measurement of bowleg deformity: variability due to method and limb rotation. J Pediatr Orthop 1994;14:147–51.
[21] Dettling S, Weiner DS. Management of bow legs in children: a primary care protocol. J Fam Pract 2017;66:E1–6.
[22] Melhem E, Assi A, El Rachkidi R, et al. EOS((R)) biplanar X-ray imaging: concept, developments, benefits, and limitations. J Child Orthop 2016;10:1–4.
[23] Chiron P, Demoulin L, Wytrykowski K, et al. Radiation dose and magnification in pelvic X-ray: EOS imaging system versus plain radiographs. Orthop Traumatol Surg Res 2017;103:1155–9.
[24] Faria R, McKenna C, Wade R, et al. The EOS 2D/3D X-ray imaging system: a cost-effectiveness analysis quantifying the health benefits from reduced radiation exposure. Eur J Radiol 2013;82:e342–9.
[25] Guven A, Hancili S, Kuru LI. Obesity and increasing rate of infantile Blount disease. Clin Pediatr (Phila) 2014;53:539–43.
[26] Rivero SM, Zhao C, Sabharwal S. Are patient demographics different for early-onset and late-onset Blount disease? Results based on meta-analysis. J Pediatr Orthop B 2015;24:515–20.
[27] Sabharwal S, Zhao C, McClemens E. Correlation of body mass index and radiographic deformities in children with Blount disease. J Bone Joint Surg Am 2007;89:1275–83.