Joint Diseases and Related Surgery

Jt Dis Relat Surg
2022;33(3):567-573

ORIGINAL ARTICLE

Evolution of the angular deformity and limb length discrepancy of congenital posteromedial bowing of the tibia over time

Ahmet Sevencan, MD, Hanifi Ucpunar, MD, Hakan Akgun, MD, Osman Nuri Ozyalvac, MD, Evren Akpinar, MD, Avni Ilhan Bayhan, MD, Timur Yildirim, MD

Department of Orthopedics and Traumatology, Metin Sabancı Baltalimani Bone Diseases Training and Research Hospital, Istanbul, Türkiye

ABSTRACT

Objectives: This study aims to assess the natural history of congenital posteromedial bowing of the tibia (CPMBT) deformity during growth and to evaluate the outcomes of lengthening by an Ilizarov frame in CPMBT patients with limb length discrepancy (LLD).

Patients and methods: Between January 2000 and December 2019, a total of 22 patients (12 males, 10 females; mean age: 10.5±4.4 years; range, 6 to 19 years) with the diagnosis of CPMBT and followed closely from birth until skeletal maturity were retrospectively analyzed. The radiographic evaluation included the anteroposterior and lateral interphyseal angle and full leg standing radiographs. Limb lengthening by an Ilizarov frame was performed for an estimated LLD over 40 mm.

Results: The age of the patients ranged from six days to 10 months at the time of presentation, while the mean age at the final follow-up was 10.5±4.4 years. Posterior medial bowing was satisfactorily remodeled in 13 (59%) patients those were not operated either for deformity or LLD. The mean LLD was 21±4.1 mm in 13 non-operated CPMBT patients. Nine of 22 (41%) patients underwent lengthening for LLD. Five of the nine CPMBT patients were operated under the age of 10 years, while four were operated over the age of 10 years.

Conclusion: According to the results of the current study, CPMBT was satisfactorily remodeled in more than half of the patients, and the majority of patients did not undergo surgery for angular deformity and LLD within 10 years of their lives. Based on these findings, although most of the patients' deformities remodeled, it should be kept in mind that some may require lengthening surgery.

Keywords: Congenital anomalies, limb lengthening; limb length discrepancy, pediatric, posteromedial bowing, tibia.

Congenital posteromedial bowing of the tibia (CPMBT) is a very rare birth defect which was firstly described by Heyman and Herndon in 1949.[1] Considering its potential to remodel, it can be accepted as a benign, self-solving, and single oblique deformity.[2-4] In the literature, it has been reported that remodeling may remain incomplete, and limb length discrepancy (LLD) increases with age which can be up to 7 cm at skeletal maturity.[5] A previous study has shown that the amount of both posterior and medial tibial bowing present at birth is positively correlated with LLD at maturity.[4] Another study has reported that the degree of tibial shortening is associated with the degree of medial bowing, but not with posterior bowing.[6]

Although there are several studies reporting that LLD increases mostly in the first year of life of a child with CPMBT, the course of LLD still remains unclear.[4-6] Therefore, the timing of surgery in children requiring surgery due to LLD has not yet been elucidated in the literature. In the current study, we hypothesized that, in patients with CPMBT deformity, a cut-off time could be determined by observing the correction of the deformity over time.

Received: May 16, 2022
Accepted: August 08, 2022
Published online: October 06, 2022

Correspondence: Ahmet Sevencan, MD. Metin Sabancı Baltalimanı Kemik Hastalıkları Eğitim ve Araştırma Hastanesi Ortopedi ve Travmatoloji Kliniği, 34470 Sarıyer, İstanbul, Türkiye.

E-mail: ahmetsevencan@hotmail.com

Doi: 10.52312/jdrs.2022.718

Citation: Sevencan A, Ucpunar H, Akgun H, Ozyalvac ON, Akpinar E, Bayhan AI, et al. Evolution of the angular deformity and limb length discrepancy of congenital posteromedial bowing of the tibia over time. Jt Dis Relat Surg 2022;33(3):567-573.

We, therefore, aimed to assess the natural history of CPMBT deformity during growth and to evaluate the outcomes of lengthening by an Ilizarov frame in CPMBT patients with LLD.

PATIENTS AND METHODS

Study population

This single-center, retrospective study was conducted at Metin Sabancı Baltalimani Bone Diseases Training and Research Hospital, Department of Orthopedics and Traumatology between January 2000 and December 2019. Patients who were firstly diagnosed with CPMBT from birth and followed only in our center were included in this study. Exclusion criteria were inappropriate radiographic data for the measurement, initial evaluation being conducted in an external center, and lost to follow-up. A total of 38 patients with CPMBT were identified; however, only 25 were referred to our hospital within the first year of age and were then closely followed. Three patients were excluded, as their radiographic data were not suitable for the measurement. Finally, a total of 22 patients (12 males, 10 females; mean age: 10.5±4.4 years; range, 6 to 19 years) with the diagnosis of CPMBT and followed closely from birth until skeletal maturity were included.

Radiographic evaluation

The following variables were measured on serial sequential radiographs to assess the initial deformity and spontaneous remodeling:

1. The anteroposterior (AP) interphyseal angle (AP-IPA) and the lateral interphyseal angle (Lat-IPA), measured between a line perpendicular to the proximal physis and a line perpendicular to the distal physis on the true AP and lateral views of the leg. The AP-IPA indicates medial bowing and Lat-IPA shows posterior bowing (Figure 1).[5-7]
2. Limb length discrepancy measured on lower limb orthoroentgenogram (LLOs). The difference compared with the contralateral side was expressed in LLD-mm.[5-7]

The serial sequential radiographs of the patients were analyzed in detail to examine the evolution of the CPMBT deformity. The change in the deformity was recorded as an angular degree at the first presentation (within the first year) and at one, two, four, five, and 10 years of follow-up (Figure 2).

Preoperative evaluation

The Green-Anderson (growth remaining) method and the multiplier method were used to estimate LLD.[8,9] Leg lengthening with an Ilizarov circular external fixator was applied to the patients with a predicted or existing length difference of more than 40 mm.

Surgical technique

The fibula was osteotomized at the proximal and middle one-third junction of the fibula.[10] Following fibular osteotomy, a previously prepared circular external fixator with three or five rings was placed on the extremity.[10] Transverse reference Kirschner wires (K-wire) were sent from lateral to medial in the metaphyseal bone, proximally parallel to the knee joint and distally parallel to the ankle joint.[10] Additional K-wires and Schanz pins were, then, placed to complete the fixation. The external fixator was destabilized to allow working on the proposed osteotomy line. In patients without an angular deformity (n=3), corticotomy was performed at 2 to 3 cm distal to the proximal fixation using multiple drill holes and osteotomy, and the external fixator was, then, stabilized.[10,11] In patients with an angular deformity (n=6), corticotomy was performed at the center of rotation and angulation of the diaphyseal deformity.[10,11] The deformity was corrected acutely and the circular external fixator was stabilized. Lengthening was performed with bifocal osteotomy in two patients (Figure 3).

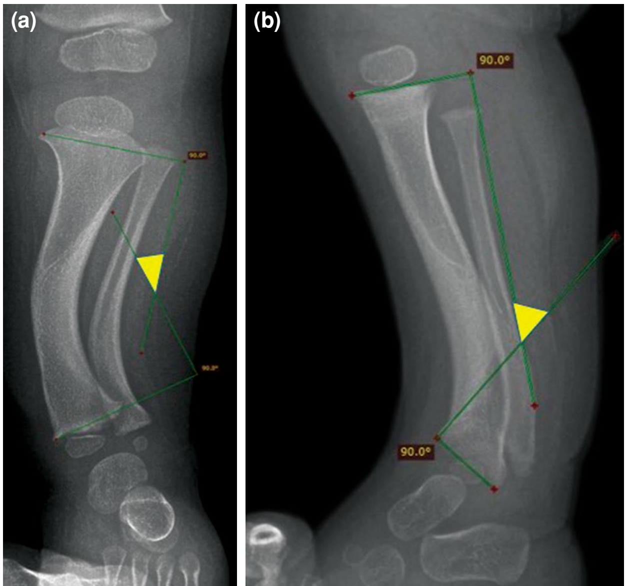

FIGURE 1. (a) Anteroposterior interphyseal angle (AP-IPA) and **(b)** lateral interphyseal angle (Lat-IPA), measured between a line perpendicular to the proximal physis and a line perpendicular to the distal physis on the true anteroposterior and lateral views of the leg. AP-IPA indicates medial bowing and Lat-IPA shows posterior bowing.

FIGURE 2. Follow-up radiographs of a patient with congenital posteromedial bowing of the tibia between the ages of 0 and 4 years. **(a)** Newborn anteroposterior interphyseal angle (AP-IPA), **(b)** first-year AP-IPA, **(c)** second-year AP-IPA, **(d)** fourth-year AP-IPA, **(e)** newborn lateral IPA (Lat-IPA), **(f)** First-year Lat-IPA, **(g)** second-year Lat-IPA, **(h)** fourth-year Lat-IPA, and **(i)** fourth-year lower limb orthoroentgenogram.

Postoperative follow-up

Active and passive knee and ankle range of motion (ROM) exercises were initiated postoperatively. The extremities were allowed to bear partial weight-bearing under pain control. Distraction was started on the third postoperative day. During the correction and distraction periods, all patients were monitored with radiographs every two weeks. They were, then, followed on a monthly basis using radiographs until full consolidation. The external fixator was retained, until consolidation was achieved.

Statistical analysis

Statistical analysis was performed using the IBM SPSS version 19.5 software (IBM Corp., Armonk, NY, USA). Descriptive data were expressed in mean ± standard deviation (SD), median (min-max) or number and frequency, where applicable. The radiographic measurements at different time points (baseline, at one, two, four, five, and 10 years) were compared using the non-parametric Wilcoxon test. A *p* value of <0.05 was considered statistically significant.

RESULTS

The demographics of the patients are shown in Table I. The age of the patients ranged from six days to 10 months at the time of presentation, while the mean age at the final follow-up was 10.5±4.4 years. The mean follow-up was 10.5±4.4 (range, 6 to 19) years. Seventeen of 22 patients (77%) did not undergo surgery for angular deformity and LLD within 10 years of their lives. In these patients, the mean initial medial bowing was 52.4±4.7 (range, 46 to 60) degrees and the mean initial posterior bowing was 47.4±10.2 (range, 38 to 61) degrees. The mean initial LLD was 10.6±4.2 (range, 7 to 18) mm. The degree of deformity in coronal and sagittal plane significantly decreased in each time point (1st *vs.* 2nd year, 2nd *vs.* 4th year, 4th *vs.* 5th year, and 5th *vs.* 10th year) during follow-up. The length of the LLD also increased

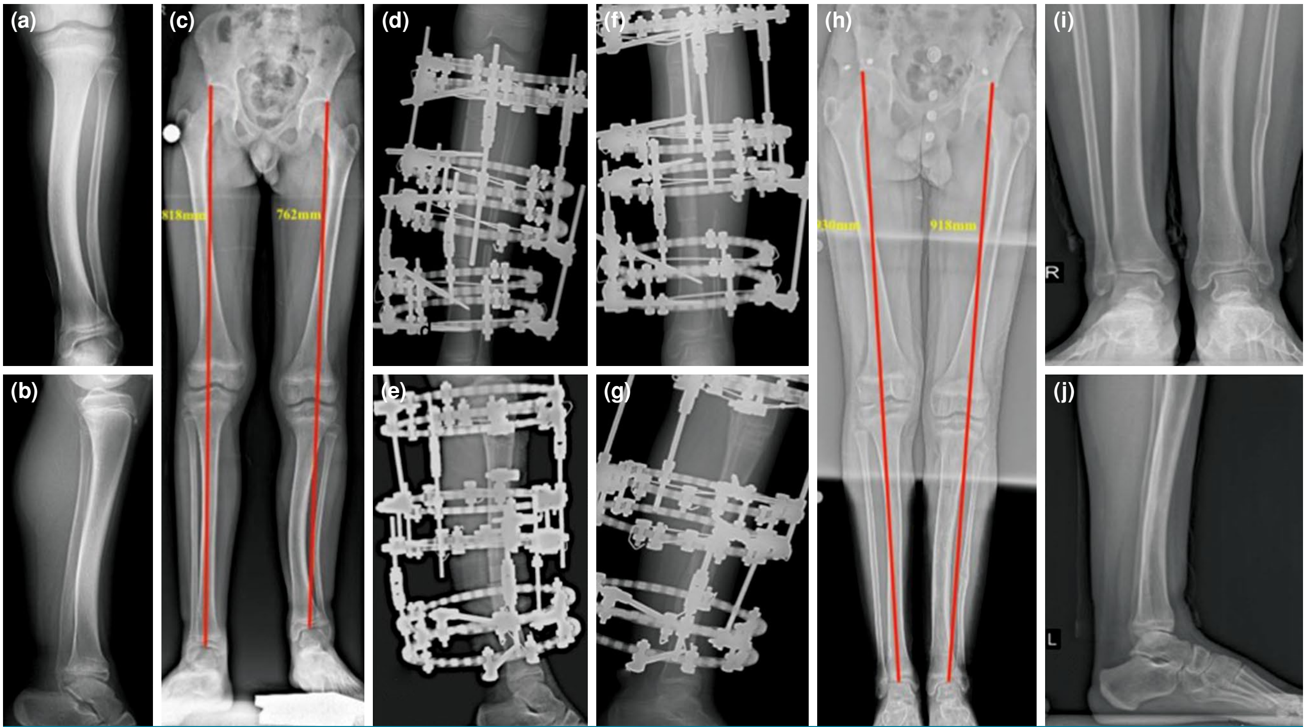

FIGURE 3. Follow-up radiographs of a patient who underwent lengthening with bifocal osteotomy. **(a)** Preoperative anteroposterior (AP) radiograph, **(b)** preoperative lateral radiograph, **(c)** preoperative lower limb orthoroentgenogram, **(d)** postoperative first-day AP radiograph, **(e)** postoperative first-day lateral radiograph, **(f)** AP radiograph after consolidation occurred, **(g)** lateral radiograph after consolidation occurred, **(h)** postoperative third-year lower limb orthoroentgenogram, **(i)** postoperative third-year ankle AP radiograph, and **(j)** postoperative third-year AP radiograph of the ankle.

significantly in each time point and the femur did not contribute to LLD (Table II, Figure 4).

Posterior medial bowing was satisfactorily remodeled in 13 of 22 (59%) patients who were not operated for deformity and/or LLD. The mean LLD of 13 non-operated CPMBT patients was 21±4.1 (range, 13 to 27) mm. Nine of 22 (41%) patients underwent lengthening with an Ilizarov circular external fixator for LLD (Table III). Five of the nine CPMBT patients were operated under the age of 10 years, while four of them were operated over the age of 10 years. Pin tract infections developed in three patients and treated with local antibiotics. Knee flexion contracture was observed in three patients and the contracture of the ankle joint in six patients, which were all treated with physiotherapy without the need for closed manipulation or open surgery.

TABLE I
Demographic data of patients (n=22)

Characteristic	n	Mean±SD	Range
Age at the last control (year)		10.5±4.4	6-19
Sex			
Female	10		
Male	12		
Side			
Left	11		
Right	11		
During follow-up (year)		10.5±4.4	6-19
LLD surgery			
Yes	9		
No	13		

SD: Standard deviation; LLD: Leg length discrepancy.

DISCUSSION

The present study was conducted with patients with CPMBT referred to a single center within the first year of life and followed regularly on a regular basis. Therefore, it provides valuable information about the course of this deformity and the development of LLD in these patients, particularly in the first 10 years of life.[12] Our study results showed that LLD was the main determinant of the need for surgical treatment in these patients and angular deformities indirectly contributed to this need. Since the growth axis of the bone is different from the

TABLE II

Follow-up radiographic measurements of the patient who did not undergo surgery within 10 years of life

	Initial		1st year		Initial vs. 1st year*	2nd year		1st year vs. 2nd year*	4th year		2nd year vs. 4th year*	5th year		4th year vs. 5th year*	10th year		5th year vs. 10th year*
	Mean±SD	Range	Mean±SD	Range	*p*	Mean±SD	Range	*p*	Mean±SD	Range	*p*	Mean±SD	Range	*p*	Mean±SD	Range	*p*
AP interphyseal angle	52.4±4.7	46-60	35.7±6.2	28-47	0.005	19.1±4.7	15-26	0.011	13.5±8.7	9-41	0.011	8.6±3.2	4-15	0.007	6.5±3.7	0-14	0.004
Lateral interphyseal angle	47.4±10.2	38-61	31.8±6.7	21-42	0.005	22.2±6.7	13-29	0.011	16.8±14.5	7-59	0.011	9.3±8.9	0-30	0.002	2.9±2.8	0-8	0.005
LLD	10.6±4.2	7-18	19.3±5.2	14-28	0.005	28±6.2	20-37	0.011	31.4±9.2	20-38	0.011	34.1±10.6	20-36	0.032	34.1±10.6	20-36	0.003

SD: Standard deviation; AP: Anteroposterior; LLD: Limp length discrepancy; * p values according to the Wilcoxon test.

TABLE III

Variables of patients undergoing lengthening surgery in our clinic

Case ID	Current age (year)	Age at operation (year)	Sex	Side	Follow-up period (year)	Deformity correction (+/-)	L-IFA		AP-IFA		OPA*	LLD	
							Before surgery (degrees)	After Surgery (degrees)	Before surgery (degrees)	After surgery (degrees)	Before Surgery (degrees)	Before surgery (mm)	After surgery (mm)
1	8	5	Male	Right	8	yes	10	0	22	0	24	48	3
2	6	4	Male	Right	5	yes	2	0	20	3	20	40	2
3	7	6	Female	Left	7	yes	12	0	14	0	18	54	0
4	6	5	Female	Right	5	yes	3	0	12	0	12	33	0
5	14	10	Male	Left	14	yes	6	0	14	4	16	56	10
6	19	13	Male	Left	19	yes	0	0	5	0	-	65	5
7	12	10	Female	Right	12	no	3	0	7	0	-	48	4
8	13	11	Female	Right	13	no	5	0	8	0	10	50	0
9	10	9	Male	Right	10	no	8	0	9	0	12	50	3

L-IFA: Final lateral interphyseal angle before lengthening; AP-IFA: Final anteroposterior interphyseal angle before lengthening, mm; millimeters; OPA: Oblique plane angle; LLD: Limb-lenght discrepancy; * Oblique plane deformity was determined using the graph method.[17]

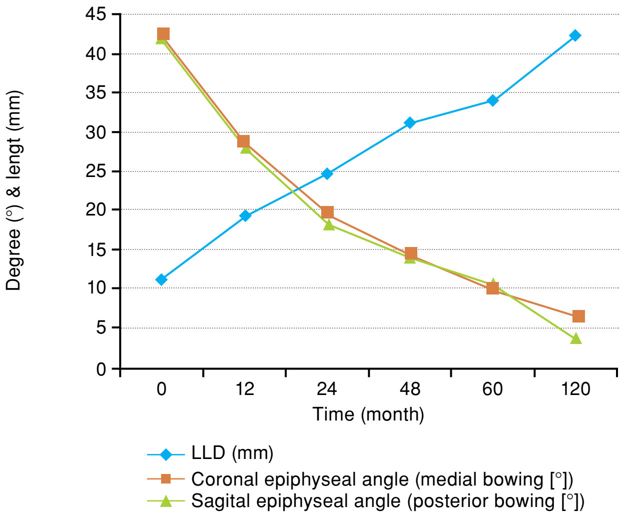

FIGURE 4. Graph showing an increase in LLD despite the tendency of angular deformity to improve over time. The measurement values of 17 patients who continued their current follow-up and did not have surgery for angular deformity and LLD in the first 10 years of their lives can be seen.
LLD: Limb length discrepancy.

neutral axis due to the angulation or bowing of the tibia, it may cause shorter bone length in the coronal plane compared to the intact tibia. However, this proposition should be further discussed and investigated in the literature, since patients whose angular deformity has completely recovered may also have short stature.

The findings described above are in line with previous studies investigating the course of the CPMBT deformity in children.[5-7,13] The degree of posterior bowing improvement was parallel to that of medial bowing in all follow-ups. This is consistent with the study conducted by Wright et al.[5] and supports previous studies suggesting that the greater the degree of bowing in the first year of life, the greater the LLD that would develop.[5-7] The rate of increase in LLD was greatest in the first year of life, when bowing was mostly improved. The reason for the high increase in the amount of leg length discrepancy in the first one-year follow-up is probably due to high the growth rate in the first two years of life. The high increase in LLD in the first year of life is consistent with previous studies.[5] The rate of increase in LLD between the two lower extremities remained constant, gradually decreasing during the first four years of life. In the following years, the increase in LLD continued, although it remained stable. This is consistent with a congenital type of (Type 1) LLD described by Shapiro,[14] as cited by Wright et al.[5] in their publication.

The timing of surgical treatment to correct deformity or height inequality in CPMBT is still controversial. Many authors recommend that surgical treatment should be performed after skeletal maturity in these patients due to the frequency of complications after early deformity surgery and the recurrence of LLD.[5-7,15,16] In a series of 20 patients, Shah et al.[6] performed lengthening before skeletal maturation in nine patients and reported several complications. Johari et al.[15] also argued that, in a series of nine patients, early surgery was associated with complications such as non-union, growth arrest, ankle valgus deformity, and LLD recurrence and, therefore, it should be avoided. Wright et al.,[5] who published the results of leg lengthening surgery in 17 patients, reported that early surgery resulted in recurrence, which then required additional epiphysiodesis and lengthening surgery. In a study conducted with 27 patients, Di Gennaro et al.[7] suggested that deformity correction could be performed at an early age, but it should be waited until skeletal maturity for lengthening. In a series of four cases with an average age of less than five years, Napiontek and Shadi[16] only corrected deformity and argued that the degree of deformity was more important for surgical timing than surgical age. Sagade et al.,[17] in their series of 23 cases in which they performed lengthening with the Ilizarov external fixator, suggested that early lengthening resulted in fewer complications and took less time. Moreover, they argued that if shortening recurred in patients who underwent early lengthening, reoperation would cause fewer complications, since less lengthening would be required. As a result of our study, we found that most of the deformities in patients with CPMBT improved within the first five years, and 77% of patients did not undergo surgery until the age of 10 years. However, it is difficult to identify the ideal age for surgery to correct LLD. We believe that postponing surgical treatment until skeletal maturity is an option in patients with relatively less residual coronal and sagittal deformities and LLD of less than 4 cm. Thus, even if a reoperation is required for LLD at an age close to maturity, the operation to be performed may cause less complications.[17]

Nonetheless, there are certain limitations to this study. First, it includes a limited number of patients, which may have skewed the data. Second, patients with severe deformities may have been referred to our center; therefore, angular deformity findings and LLD severity could vary. In particular, cases with mild posteromedial deformities may not have been referred to our center, potentially leading to an overestimation of the LLD frequency and rate.

In conclusion, according to the results of the current study, CPMBT was satisfactorily remodeled in more than half of the patients, and the majority of patients did not undergo surgery for angular deformity and LLD within 10 years of their lives. Based on these findings, although most of the patients' deformities remodeled, it should be kept in mind that some may require lengthening surgery.

Ethics Committee Approval: The study protocol was approved by the Metin Sabancı Baltalimani Bone Diseases Training and Research Hospital Ethics Committee (date: 14.04.2021, no: 72/505). The study was conducted in accordance with the principles of the Declaration of Helsinki.

Patient Consent for Publication: A written informed consent was obtained from each patient.

Data Sharing Statement: The data that support the findings of this study are available from the corresponding author upon reasonable request.

Author Contributions: Material preparation, data collection and analysis were performed: A.S., H.A., E.A., A.I.B., O.N.O. and T.Y.; The first draft of the manuscript was written by: A.S., and H.U.; All authors contributed to the study conception and design. All authors commented on previous versions of the manuscript. All authors read and approved the final manuscript.

Conflict of Interest: The authors declared no conflicts of interest with respect to the authorship and/or publication of this article.

Funding: The authors received no financial support for the research and/or authorship of this article.

REFERENCES

1. Heyman CH, Herndon CH. Congenital posterior angulation of the tibia. J Bone Joint Surg [Am] 1949;31:571-80.
2. Rathgeb JM, Ramsey PL, Cowell HR. Congenital kyphoscoliosis of the tibia. Clin Orthop Relat Res 1974;(103):178-90.
3. Hill R. Congenital Posteromedial Bowing of the Tibia (Congenital Tibia Recurvatum). In: Rozbruch SR, Hamdy R, editors. Limb Lengthening and Reconstruction Surgery Case Atlas. Berlin: Springer; 2014. p. 1-5.
4. Pappas AM. Congenital posteromedial bowing of the tibia and fibula. J Pediatr Orthop 1984;4:525-31.
5. Wright J, Hill RA, Eastwood DM, Hashemi-Nejad A, Calder P, Tennant S. Posteromedial bowing of the tibia: A benign condition or a case for limb reconstruction? J Child Orthop 2018;12:187-96.
6. Shah HH, Doddabasappa SN, Joseph B. Congenital posteromedial bowing of the tibia: A retrospective analysis of growth abnormalities in the leg. J Pediatr Orthop B 2009;18:120-8.
7. Di Gennaro GL, Gallone G, Martinez Vazquez EA, Marchesini Reggiani L, Racano C, Olivotto E, et al. Deformity progression in congenital posteromedial bowing of the tibia: A report of 44 cases. BMC Musculoskelet Disord 2020;21:430.
8. Anderson M, Green WT, Messner MB. Growth and predictions of growth in the lower extremities. J Bone Joint Surg [Am] 1963;45:1-14.
9. Paley D, Bhave A, Herzenberg JE, Bowen JR. Multiplier method for predicting limb-length discrepancy. J Bone Joint Surg [Am] 2000;82:1432-46.
10. Schoenecker PL, Gordon JE. Operative treatment of tibia vara. In: Tolo VT, Skaggs DL, editors. Master techniques in orthopaedic surgery: pediatrics. Philadelphia: Lippincott Williams & Wilkins; 2008. p. 297-302.
11. Gordon JE, Schoenecker PL, Lewis TR, Miller ML. Limb lengthening in the treatment of posteromedial bowing of the tibia. J Child Orthop 2020;14:480-7.
12. Atik OŞ. What are the expectations of an editor from a scientific article? Jt Dis Relat Surg 2020;31:597-8.
13. Kaufman SD, Fagg JA, Jones S, Bell MJ, Saleh M, Fernandes JA. Limb lengthening in congenital posteromedial bow of the tibia. Strategies Trauma Limb Reconstr 2012;7:147-53.
14. Shapiro F. Developmental patterns in lower-extremity length discrepancies. J Bone Joint Surg [Am] 1982;64:639-51.
15. Johari AN, Dhawale AA, Salaskar A, Aroojis AJ. Congenital postero-medial bowing of the tibia and fibula: Is early surgery worthwhile? J Pediatr Orthop B 2010;19:479-86.
16. Napiontek M, Shadi M. Congenital posteromedial bowing of the tibia and fibula: Treatment option by multilevel osteotomy. J Pediatr Orthop B 2014;23:130-4.
17. Sagade B, Jagani N, Chaudhary I, Chaudhary M. Congenital posteromedial bowing of tibia: Comparison of early and late lengthening. J Pediatr Orthop 2021;41:e816-e822.