

## Case Reports and Series

## Correction of Longitudinal Epiphyseal Bracket Disease with External Fixation: A Case Report with 6-year Follow-up Period

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## ABSTRACT

Longitudinal epiphyseal bracket disease is a rare congenital abnormality of the tubular bones. This syndrome eventually leads to progressive shortening and angular deformity of the involved bone. When longitudinal epiphyseal bracket disease affects the first metatarsal, a short trapezoidal shaped metatarsal is noted radiographically. We present the case of a 9-year-old patient with longitudinal epiphyseal bracket disease of the first metatarsal. The abnormality was surgically treated by bracket osteotomy with callus distraction before physeal closure. At 6 years of follow-up, the patient displayed a relatively normal looking cosmetic foot compared with the contralateral foot. A mild residual hallux varus was noted, with maintenance of the forefoot parabola. Surgical correction resulted in alteration of the first intermetatarsal angle and the first metatarsophalangeal angle. The first intermetatarsal angle decreased from a preoperative value of  $-30^\circ$  to a postoperative value of  $10^\circ$ . The first metatarsophalangeal angle decreased from a preoperative value of  $36^\circ$  of hallux varus to a postoperative value of  $4^\circ$ . The length of the first metatarsal was restored compared with that of the contralateral limb and was slightly elongated. The present case study demonstrates that early surgical correction with callus distraction of an ossified abnormal bracket before physeal closure, a relatively simple procedure, can restore longitudinal growth and correct the angular deformity.

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Longitudinal epiphyseal bracket disease (LEBD), also referred to as *delta bone* or *delta phalanx* is a rare congenital ossification anomaly (1–3). LEBD is initially composed of cartilage forming a bracket linking the proximal and distal epiphyses of the tubular bones and causing defective growth of the primary ossification focus. Changes in bone growth secondary to the presence of LEBD would cause angular malformations and length discrepancies (4). This anomaly is most often recognized in the hands and feet at the levels of the phalanges, metacarpals, and metatarsals. When it affects the first metatarsal, the longitudinal metatarsal epiphyseal bracket leads to a short, broad, trapezoidal metatarsal with deviation of the metatarsophalangeal joint and hallux varus. According to a literature review, LEBD has been associated with numerous syndromes, including Rubinstein-Taybi syndrome, Cenani-Lenz syndactyly, isolated oligosyndactyly, and Nievergelt syndrome (5). Multiple treatment options are available, including excision of the cartilaginous bracket to prevent future soft tissue contractures and osseous deformities, bone grafting, and bone

transport using external fixation (3,6,7). The purpose of the present study is to report the long-term follow-up of a patient with LEBD treated with callus distraction.

### Case Report

A 9-year-old girl presented to the senior author's (C.K.) office complaining of a right foot congenital deformity. She reported exquisite pain along the medial aspect of her right foot while dancing and was concerned with the cosmesis of her foot. A small joint effusion was noted sub second during physical exam which was extremely tender to palpation. The history and physical examination revealed no underlying medical conditions associated with LEBD. The initial radiographic examination showed brachymetatarsia of the right first metatarsal with lateral deviation of the metatarsal phalangeal articulation (Fig. 1). Both surgical and conservative interventions were discussed with the patient and her family. Surgical intervention was chosen by all involved.

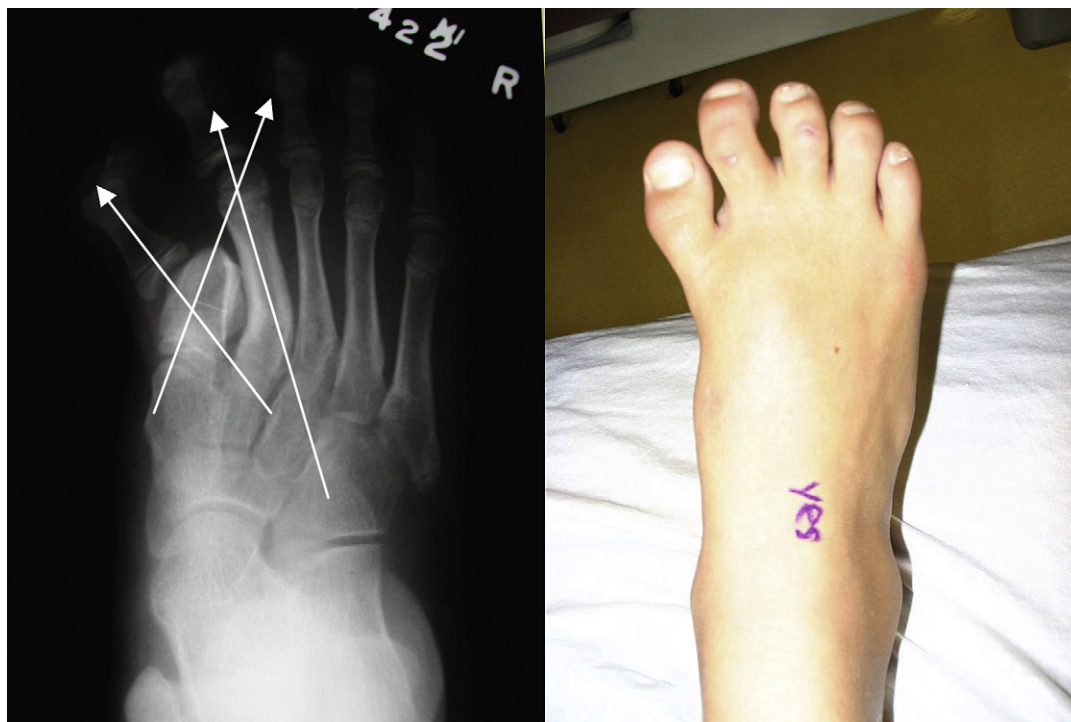
All surgical interventions were performed as outpatient procedures with the patient under general anesthesia. The initial surgical intervention performed included a transverse osteotomy of the first metatarsal in the metaphyseal bone with application of a minirail

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**Fig. 1.** Preoperative radiographic and clinical images demonstrating longitudinal epiphyseal bracket disease.

external fixator (OrthoFix International, Verona, Italy) and an oblique osteotomy with Kirschner wire fixation of the second metatarsal. Excision of the cartilaginous “bracket” was also performed during the initial surgical procedure. The external fixator pins were placed dorsally in the medial cuneiform and the base of the first metatarsal proximally and dorsally in the distal shaft of the first metatarsal. Callus distraction of the first metatarsal using the external fixator followed the standard distraction principle of 1 mm/day. Initial compression of the osteotomy was performed for 7 days, followed by gradual distraction of the first metatarsal at 1 mm/day. Distraction was performed until the minirail had reached its maximum distraction length. The patient then underwent removal of the minirail, with reapplication and modification of pin placement for additional correction of the first metatarsal. The proximal pins were left in their original position and the distal pins of the external fixator were placed in the head of the first metatarsal and the base of the proximal phalanx of the hallux. The second minirail external fixator (OrthoFix International; Fig. 2) allowed for plantarflexion and abduction of the distal first metatarsal by way of an appropriately placed hinge over the metaphyseal diaphyseal junction. Distraction continued until the proposed radiographic length was achieved compared with the contralateral foot. The patient was then placed in a short leg fiberglass cast with the foot placed in a neutral position with a strict non-weight-bearing status for the initial 3 weeks. She was then allowed progressive weight-bearing in the fiberglass cast for 3 weeks. After removal of the fiberglass cast, the hallux was splinted to the second digit owing to a residual varus angulation of the digit. Residual hallux varus was noted several months later, with contracture of the hallux interphalangeal joint right foot. She returned to the operating room for soft tissue release, lateral capsulotomy, and first metatarsal Chevron osteotomy with screw fixation (OsteoMed Orthopaedics, Dallas, TX). The capital fragment was rotated medially to laterally for articular cartilage realignment with the base of the proximal phalanx. Lateral capsulorrhaphy was

performed to further correct the residual hallux varus. Serial radiographs were taken throughout the entire treatment plan to ensure appropriate correction.

The patient was followed up 6 years later with physical and radiographic examinations. She was able to resume all activity, without any significant complaints. She had a decrease in the range of motion at the first metatarsophalangeal joint, with a residual 5° of dorsiflexion and 20° of plantarflexion. The patient displayed a relatively normal looking cosmetic foot compared with the contralateral foot. A mild residual hallux varus was noted, with maintenance of the forefoot parabola. Surgical correction resulted in alteration of the first intermetatarsal angle and first metatarsophalangeal angle. The first intermetatarsal angle decreased from a preoperative value of -30° to a postoperative value of 10°. The first metatarsophalangeal angle decreased from a preoperative value of 52° of hallux varus to a postoperative value of 4°. The length of the first metatarsal was restored compared with that of the contralateral limb and was slightly elongated, causing an angulation deformity of the hallux and adaptive changes in the cartilage laterally (Figs. 3 and 4). The first metatarsal was longer than that of the contralateral first metatarsal on the 6-year radiographs. We believe this might have resulted from the longitudinal growth associated maturation of the midfoot. Synostosis was noted on the anteroposterior radiograph between the first and second metatarsal, which was not planned during correction. We believe this was most likely due to the pediatric patient's ability to aggressively lay down new bone callus for correction of disruptions/fractures. To date, the synostosis has not caused any pain or discomfort to the patient; however, this does pose a possible cause for long-term angular deviation as the patient ages. Angular correction of the second metatarsal was also noted compared with the preoperative radiographs. This patient should be followed up chronically, because the ossification centers of the foot will begin to close and her true foot structure will be revealed, unmasking any potential deformities that might require additional correction.

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