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# Metatarsophalangeal Arthritis Following Fourth Metatarsal Lengthening Treated With Distraction Arthroplasty: Case Report

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**Level of Evidence:** Level V, expert opinion.

**Keywords:** brachymetatarsia, metatarsophalangeal arthritis, distraction, osteogenesis, arthroplasty

Brachymetatarsia is congenital shortening of the metatarsal (MT), which usually results in cosmetic deformities and can cause transfer metatarsalgia. The operative management of this condition involves lengthening to restore the parabolic arch of the foot as well as the length of the MT to normalize load transfer across the MT heads. Distraction osteogenesis via external fixation has been used successfully in the gradual correction of brachymetatarsia.[12,16,19] However, this method has been associated with a number of complications, including metatarsophalangeal (MTP) stiffness and arthritis,[16,17,27,29] with no clear algorithm on how to manage such complications. The aim of this case report was to present distraction arthroplasty of the MTP joint associated with autologous iliac crest bone marrow aspirate (ICBMA) injection as a successful treatment of an arthritic MTP joint that developed as a complication of distraction osteogenesis for third brachymetatarsia.

## Case Report

A 39-year-old female presented to the office of the senior author with congenital shortening of the left third and right fourth MTs with complaints of both embarrassment from the deformity and discomfort. Specifically, the patient was an active runner and complained of increased pain (transfer metatarsalgia), exacerbated by exercise, under her adjacent longer toes of approximately 1 year duration. She had tried MT pads and orthotics without significant relief of her symptoms. Clinically, the patient had normal ankle and hindfoot/midfoot alignment. Both affected MTs were shorter than the adjacent MTs (Figure 1A) and had mild dorsal subluxation of the MTPs. The MTP joint ranges of motion were approximately 40 degrees of dorsiflexion and 30 degrees of plantarflexion bilaterally and symmetrically. The proximal interphalangeal joints (PIPs) of both affected rays revealed no abnormalities. Examination of adjacent MTPs revealed metatarsalgia with moderate callosities.

Weight-bearing AP and lateral radiographs of both feet revealed a MT deficiency of 18 mm on the left third MT and 12 mm on the right fourth MT (Figures 1B and 1C) representing 40.9% and 27.3% shortening, respectively. The desired length was determined by using the second MT as the focal point for an arc drawn on the radiograph establishing the MT parabola, then determining the length needed from the distal aspect of the affected MT to reach the parabola on each foot (Figures 1B and 1C).[12,31]

The patient underwent staged operative correction with distraction osteogenesis of the left brachymetatarsia followed 6 weeks later by the right side. A mini-monolateral external fixator frame was applied to the dorsal aspect of the MT parallel to the longitudinal axis of the MT in both the coronal and sagittal planes. A MT osteotomy was then performed percutaneously[13,17] and the MTP joints were stabilized by temporarily spanning 0.045 K-wires driven from the distal phalanx proximally into the MTP joint (Figure 2). These wires were removed 6 weeks after the initial surgery.[31] Postoperatively, the patient was allowed heel weight-bearing and started distraction after a latency period of 10

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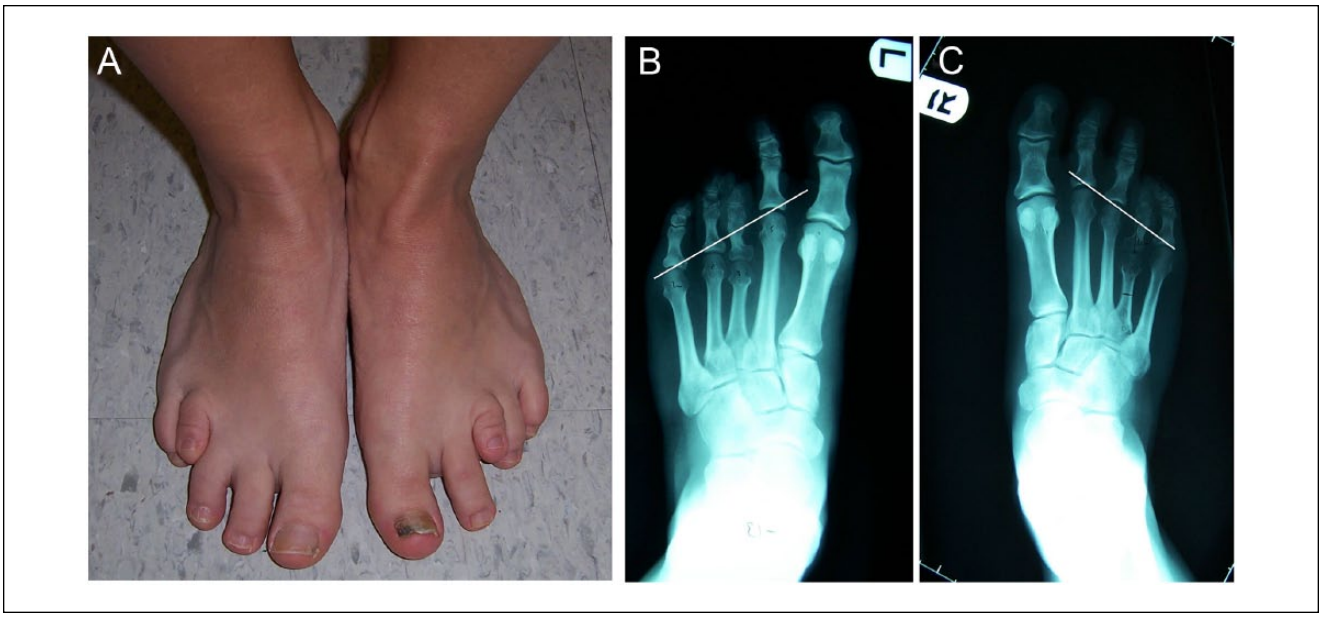


**Figure 1.** Preoperative clinical photograph and radiographs (A) Clinical photographic image of the patient's feet showing the congenital brachymetatarsia of the left third and right fourth metatarsals. (B,C) Standing frontal radiographs of both feet demonstrating shortening of the left third metatarsal by 18 mm (40.9%) and the right fourth metatarsal by 12 mm (27.3%) calculated by measuring the distance from the short metatarsal head to the line defining the metatarsal head parabola (white line) extending from the second to the fifth MTP joints.

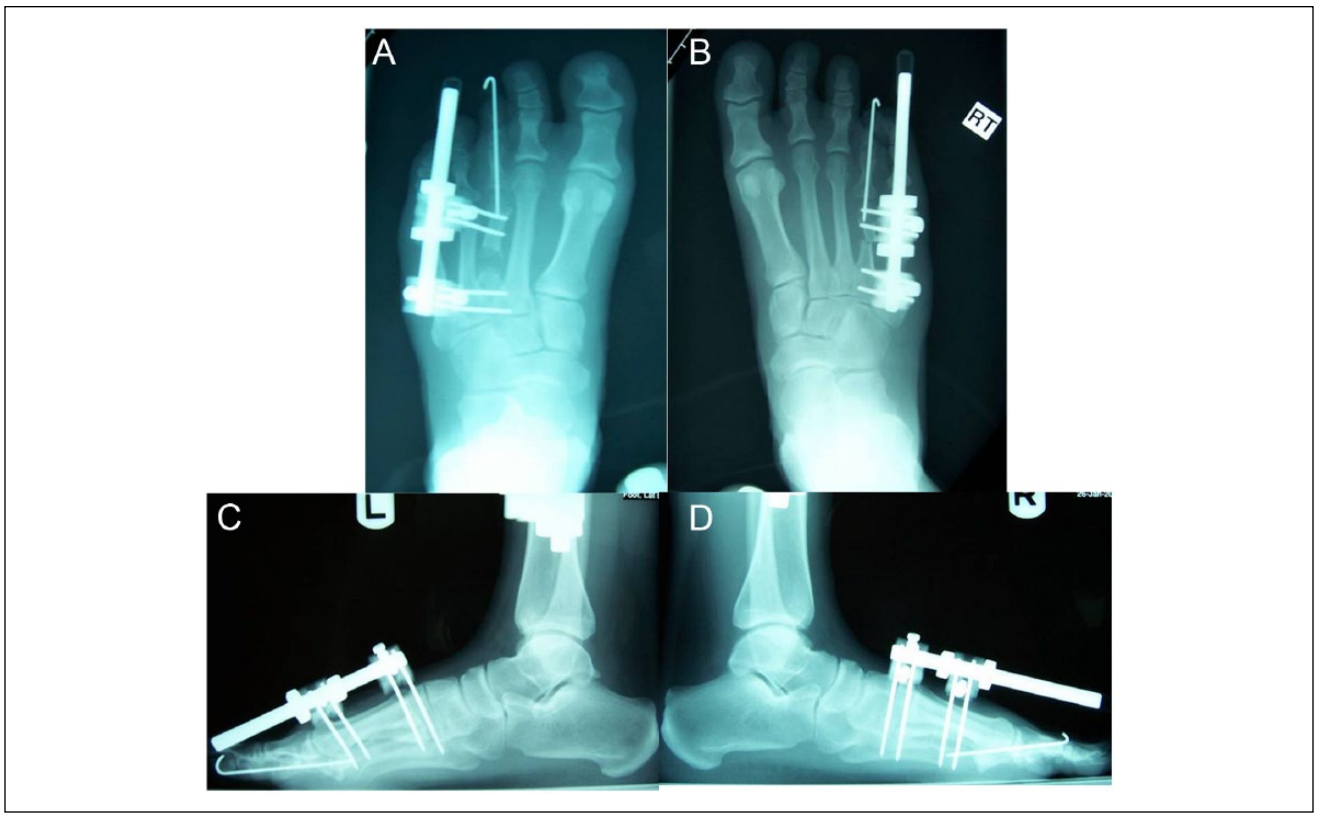


**Figure 2.** Postoperative (A-B) frontal radiographs and (C-D) lateral radiographs of both feet after external fixator application 1 week postoperatively on the right side and 4 weeks postoperatively on the left side with MTP fixation with a Kirschner-wire. Note progression of distraction gap on left third metatarsal at 4 weeks postoperatively.

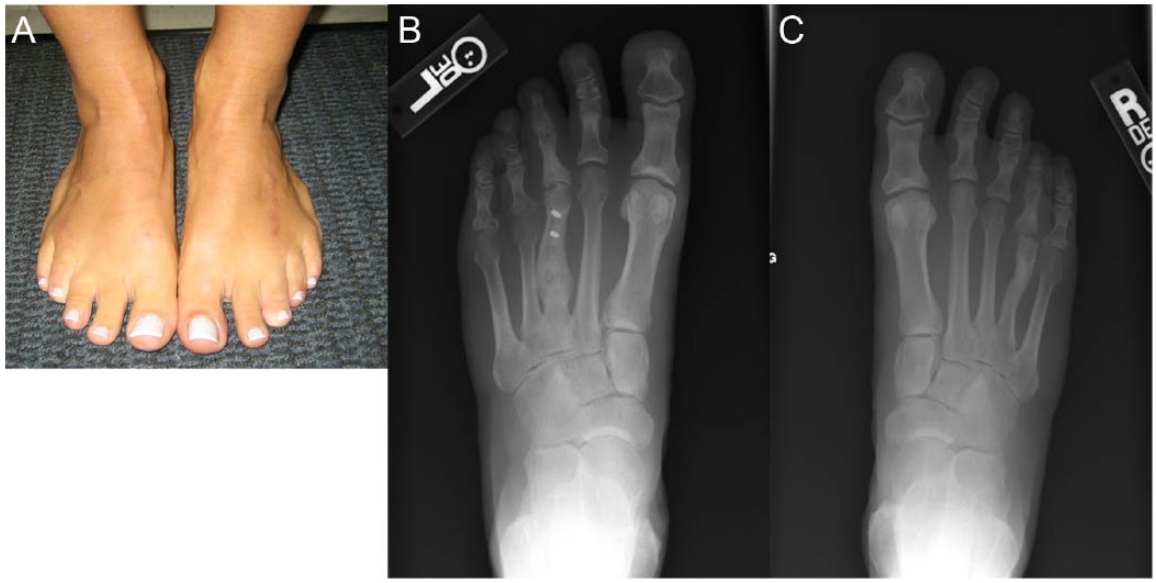


**Figure 3.** (A) Clinical photographic image and (B-C) AP radiographs of both feet 1 year postoperatively after final operative corrections showing complete consolidation of bony regenerate and restoration of lengths of the left third and right fourth metatarsals to fall on the toe-tip parabolic arches of both feet. Early degenerative changes are noted in both MTP joints. Note the 2 small metal objects on the left third MT; these represent the 2 most distal half pins of the frame that had broken toward the end of the consolidation phase with advancement of weight-bearing on the left foot. The broken pin tips were asymptomatic and therefore were left in situ after frame removal.

days at a rate of 0.5 mm/day, divided into 2 adjustments of 0.25 mm each[7,18,31] and followed up by biweekly radiographs.

The right brachymetatarsia proceeded well with distraction. The 13 mm gain in length was accomplished after 30 days of distraction. The spanning MTP K-wire was removed 6 weeks postoperatively and the frame was removed 94 days postoperatively, with a healing index of 62 days/cm. There was moderate stiffness of the MTP joint that was managed by passive stretching and range of motion exercises. The left brachymetatarsia, however, had a different and problematic course of progression. Six weeks postoperatively the patient complained of left foot pain and minimal swelling of the involved MT, together with some bending of the most distal pin of the frame. Radiographs revealed a distraction gap of 10 mm with good MT alignment yet with early bony consolidation of the regenerate with breakage of the most distal pin and bending of the second distal pin. A decision to halt distraction was made and the MT was allowed to achieve full consolidation with removal of the K-wire at 6 weeks and the frame at 3 months postoperatively. This was followed by a second surgery with repeat osteotomy, frame application, and MTP wire fixation that was performed 8 months from the initial surgery. Distraction was successfully achieved and the left third MT was lengthened to the originally planned total length of 18 mm. The patient was satisfied with the clinical appearance of both feet (Figure 3A). The frame was removed with a healing index of 75 days/cm. It is worth mentioning that the 2 most distal pins of the frame had broken toward the end of the consolidation phase with advancement of weight-bearing on the left foot. The broken pin tips were asymptomatic and therefore were left in situ after frame removal to avoid creating stress risers in the lengthened MT shaft through attempts at their removal (Figure 3B). The left third MTP joint exhibited similar moderate stiffness postoperatively (20 degrees of dorsiflexion and 30 degrees of plantarflexion), which was again managed by passive stretching and range of motion exercises. The patient was symptom free and returned to her activity levels and intensive training. She was able to run marathons.

Five years later, the patient presented with pain in the left third MTP aggravated by weight-bearing. Clinical examination revealed moderate swelling of the MTP joint with tenderness over the joint, palpable osteophytes and diminution of range of motion compared to postdistraction (10 degrees of dorsiflexion and 20 degrees of plantar flexion). Radiographs confirmed advanced left third MTP arthritis (Figure 4A). She was managed with an MTP intra-articular steroid injection and a MT pad that provided symptomatic


**Figure 4.** (A) PA radiograph of the left foot 5 years postoperatively demonstrating advanced third MTP arthritis with no joint space, osteophytes, and subchondral sclerosis. (B) Intraoperative PA fluoroscopic image after application of static mini-monolateral external fixator frame and distraction of the left third MTP joint. Note intra-articular injection of bone marrow aspirate concentrate. (C) Radiograph 2 years after third MTP distraction frame removal demonstrating 2 mm of maintained MTP joint space without progression of arthritic changes. (D) Coronal proton density, (E) coronal inversion recovery, and (F) thin-slice sagittal proton density MRI sequences 2 years at the same time point demonstrating maintenance of a thin layer of hyaline-like reparative tissue at the cartilage interface of the involved left third MTP joint.

relief for 6 months, after which the symptoms recurred. The patient was offered an MTP fusion but refused this option given her relatively good range of motion and high demands. The decision was then made to salvage the MTP joint by distraction arthroplasty. This was achieved by a left third MTP arthrotomy; exostectomy; microfracture via drilling of the arthritic MT head with 0.45 K-wire; application of a static mini-monolateral external fixator spanning the MTP joint, with 4 mm joint distraction; and injection of autologous ICBMA (Figure 4B). It is the most senior author's standard of care to routinely augment distraction arthroplasty with an adjunct biological approach in the form of microfracture of denuded areas of cartilage and autologous ICBMA injection. This has the potential of biologically augmenting the mechanical decompression of the joint achieved through distraction arthroplasty. Such an approach of using biologics in managing articular cartilage lesions and degeneration has been previously described in strategies for treating chondral lesions of other weight-bearing joints.[8,21,28]

Postoperatively, the patient was allowed heel weight-bearing and the frame was removed after 2 months when MTP ROM exercises were initiated. Two years post–frame removal, the patient remained completely pain free. MTP examination revealed no tenderness and ROM had improved to 30 degrees of dorsiflexion and 30 degrees of

plantarflexion. Radiographs revealed a maintained joint space of 2 mm comparative to prearthritic measurements of 0 mm (Figure 4C). MRI of the forefoot revealed maintenance of a thin layer of reparative tissue at the cartilage surface of the involved MTP joint with only minimal underlying subchondral reactive signal (Figures 4D and 4E).

## Discussion

The main goal of operative correction of brachymetatarsia is restoration of a functional MT parabola by correcting the deformity. While single-stage acute lengthening with an interpositional bone graft and plate fixation has the advantage of instant gratification, the amount of length attainable is limited. In addition, acute lengthening puts the neurovascular and musculotendinous structures of the foot at risk of injury from excessive tension.[12] Distraction osteogenesis using external fixation has revolutionized the management of brachymetatarsia.[6,11,13] The advantages of this method include the ability to achieve greater lengthening than with single-stage procedures, postoperative adjustability, the avoidance of graft donor site morbidity, the possibility for simultaneous multiple MT lengthening procedures, and a lower incidence of neurovascular damage.[3,6,12,23] While it is not recommended to *acutely* lengthen brachymetatarsia with bone grafting and internal fixation for more than 40% of the original MT length, distraction osteogenesis with external fixation allows for lengthening that requires achievement of 40% to 60% of the original MT length.[6,10-12,27]

Despite its advantages, the incidence of complications from distraction osteogenesis in brachymetatarsia ranges from 30% to 100%.[12,17,23] These complications can be classified as major or minor. Major complications include malalignment of the MT, MTP joint dislocation or arthritis, nonunion, and callus fracture. Minor complications include pin tract infection, pin breakage, MTP joint subluxation and stiffness.[11,14,20,27]

MTP joint stiffness and subsequent arthritis is a common complication seen after distraction osteogenesis. This may be due to a number of predisposing factors. The excessive lengthening during distraction creates tension in the lengthened tendons and the adjacent soft tissues traversing the joint including the surrounding capsule, transverse MT ligament, and the tendon sheath, which might exceed the limits of these structures and is disproportionate to tension remodeling in the lengthened bone. This can ultimately lead to increased MTP joint contact pressures that predisposes to arthritis. Therefore, distraction rates of 0.25 mm 2 or 3 times a day have been recommended by most authors for optimal soft tissue elongation and bone formation without MTP stiffness or arthritis development.[6,14,17] In addition, a number of studies have reported greater MTP joint pathologies in patients with a length gain of more than 40%.[2,3,12,23] It is worth highlighting that in this particular case report the left fourth MT was significantly shorter than its counterpart right fourth brachymetatarsus, subsequently requiring more lengthening. This excessive lengthening—albeit targeting to merely reach the MT head parabola without any overdue lengthening—might have contributed to the development of MTP arthritis in this case due to the reasons aforementioned.

In the most senior author's practice, tendon lengthening of the involved brachymetatarsus was not routinely performed as a prophylactic simultaneous procedure with distraction osteogenesis, but was rather reserved for patients who had significant tension on the tendons of the involved MT on initial presentation, evidenced by significant subluxation or dislocation of the involved MTP joint. Tendon lengthening was also performed during the course of MT lengthening if such complications of the MTP joint developed and was mainly carried out at the time of frame removal. Our algorithm of treatment gradually evolved to prophylactic extensor and flexor simultaneous tenotomies at the time of the MT osteotomy and frame application to avoid the undue excessive tension on the tendons during lengthening that increase the MTP joint contact pressures, which might contribute to the progression of MTP stiffness or arthritis. In this particular case, no tendon lengthening was performed either concomitantly with or during the course of distraction osteogenesis.

MTP joint arthritis can also occur secondary to MTP joint subluxation/dislocation with incongruence and altered joint contact pressures. It can also occur iatrogenically from prophylactic stabilization of the MTP joint with temporary K-wire fixation as noted above. The initial trauma by the K-wire to the intact cartilage can initiate a cascade of inflammatory events that may lead to the progression of the degenerative changes, particularly with repeated pinning attempts and generation of thermal necrosis associated with closed drilling across the joint. We no longer place a pin across the joint because of these issues. We stabilize the MTP joint during distraction by spanning the joint with the fixator.

Another important factor in the development of MTP arthritis after distraction is determined by the final position of the MT head after lengthening. Frames placed dorsally parallel to the longitudinal axis of the MT shaft in the sagittal plane—rather than parallel to the ground—aid in bringing the MT head plantarly with lengthening. While this ultimately aids in more load being borne by the head of the lengthened MT to take load off the overloaded adjacent MTs and relieve metatarsalgia, inadvertent excessive lengthening may lead to increased plantar displacement of the MT head, leading to excessive MTP joint loading, increased contact pressures, and subsequent arthritis.

Premature consolidation of the bony regenerate with distraction osteogenesis is a relatively common obstacle that is frequently encountered with lengthening by the Ilizarov

technique. It is considered an *obstacle* rather than a true complication. While a complication is a true problem during lengthening that is not resolved before the end of treatment, an obstacle is defined as a difficulty during distraction osteogenesis that required an operative intervention with complete resolution.[25] Therefore, premature consolidation of regenerate is not considered a true complication and its management by subsequent osteotomy would not be considered as revision surgery that would normally jeopardize the final outcome of distraction osteogenesis or precipitate MTP arthritis. Moreover, the patient in this case report developed MTP arthritis after a symptom-free interval of 5 years from the second surgery on the left foot, which represents a relatively long follow-up period that would have been significantly compromised if the second surgery had been a precipitating factor for the development MTP arthritis.

Distraction arthroplasty or arthrodiastasis is a relatively novel joint preserving procedure that has developed over the past few years. Basic science research of joint distraction has shown that generating intermittent fluid pressure while prohibiting mechanical loading has an effect on the healing capacity of the chondrocyte.[5] In addition, clinical studies on ankle distraction have demonstrated decreased joint reaction forces, with subsequent decompression of articular surfaces and subchondral bone remodeling,[9] pain relief and improved clinical function.[26,30] While this treatment modality has been effective in the ankle, there are very few reports on the results in MTP joints. Abraham et al[1] recently reported on the results of distraction arthroplasty for treatment of first MTP arthritis (hallux rigidus) with statistically significant reduction of pain and increased satisfaction after surgery. Another small case series by Xie et al[32] on treatment of late Frieberg's disease with dorsal wedge osteotomy and joint distraction arthroplasty reported statistically significant improvement in pain scores and MTP range of motion compared to preoperative levels.

The use of biological adjuncts such as ICBMA and PRP (platelet-rich-plasma) for cartilage repair has been reported in the literature and has been shown to promote the regeneration of cartilage repair tissue.[8,21,28] Therefore, addition of ICBMA has been routinely employed by the most senior author in distraction arthroplasty of other weight-bearing joints such as the ankle (unpublished data). While distraction arthroplasty mechanically unloads the joint for a prolonged period, we believe the addition of microfracture to areas of eburnated cartilage and injection of autologous ICBMA provides a booster milieu of mesenchymal stem cells (MSCs) that are capable of potentiating the healing response, via the release of chondrogenic growth factors as well as cellular contribution to chondrogenesis. Although the addition of ICBMA may be a confounding factor, it is hypothesized that the sustained effect of prolonged mechanical unloading of the joint through distraction arthroplasty outweighs the transient short-term effect of MSCs and their released growth factors from the ICBMA. In vivo studies have documented that the contribution of implanted stem cells and chondrocytes to the reparative process of cartilage regeneration is short-lived and does not exceed the first few weeks after implantation.[22,24] Moreover, the MTP joint space is extremely small and cannot hold more than 0.5 ml of injected ICBMA, in contrast to other larger weight-bearing joints such as the ankle. While the minimum threshold of injectable or implanted cells required for potentiating chondrogenesis has been reported to be around $1\text{-}2 \times 10^6$ cells/cm$^2$ of cartilage defect area,[4,15] it is highly unlikely that a smaller number of MSCs delivered to the MTP joint via a small-volume ICBMA injection would have an overwhelming (as opposed to a temporary synergistic) effect on the reparative tissue as compared to the prolonged distraction and mechanical unloading of the joint. Further in vivo studies are still required to unmask such confounding effect and further establish a cause–effect relationship of ICBMA injection with distraction arthroplasty.

To the authors' knowledge, this is the first case report that demonstrates short-term improvement in radiographic and MRI outcomes of the MTP joint in addition to clinical outcomes after development of arthritis as a complication of distraction osteogenesis for brachymetatarsia. At 2 years follow-up after removal of the MTP frame, the patient had maintained MTP joint space on radiographs, with a hyaline-like repair tissue and subchondral bone remodeling with minimal edema on MRI across the previously arthritic joint. This is in addition to a pain-free MTP joint with good range of motion.

## Conclusion

Although a devastating potential complication, we conclude that MTP arthritis salvage via distraction arthroplasty and autologous ICBMA is a viable option in complicated cases of brachymetatatrsia following lengthening by distraction osteogenesis. This might have potential implications for incorporating MTP distraction with injection of biological adjuncts as part of an algorithm for prevention and management of such a major complication. Further clinical studies with a larger number of patients including clinical and radiological midterm and long-term follow-up are needed to further validate this treatment.

### Declaration of Conflicting Interests

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