Distraction osteogenesis of fourth brachymetatarsia

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A B S T R A C T

Background: Brachymetatarsia is a rare congenital or developmental condition that results in a short metatarsal. The condition most commonly affects the fourth metatarsal of young and adolescent females. It does not usually produce a functional problem. However, it may produce a significant cosmetic problem especially in young women. The authors present their experience in gradual distraction of the fourth metatarsal to tackle this problem in adults.

Methods: That was done using a monolateral frame in 11 feet of female patients with an average age of 23 years. Evaluation depended upon the achievement of the target length, angulation, pain, and satisfaction of the patient.

Results: There were eight excellent and three good results after an average follow up of 2.6 years. Aside from nine cases of mild pin-tract infection, complications were few and minor.

Conclusions: We recommend distraction osteogenesis as the treatment of choice for brachymetatarsia of the fourth toe.

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1. Introduction

Brachymetatarsia is a rare foot condition characterized by a short metatarsal. Brachymetatarsia is diagnosed when one metatarsal ends 5 mm or more proximal to the parabolic arc [1]. A morphofunctional study described the angle of fourth metatarsal shortening or second-fourth angle to enable quantification of the shortening of the fourth metatarsal in brachymetatarsia. Brachymetatarsia is defined as a second-fourth angle of less than 52.2° in males and 50.5° in females [2].

The etiology is unknown and is associated with congenital and developmental causes, traumatic or iatrogenic factors, and it is also related with certain systemic conditions like pseudohypoparathyroidism, Turner’s syndrome, Down’s syndrome, Apert syndrome, and poliomyelitis [3–7]. The idiopathic congenital etiology, which refers to the early closure of the epiphyseal plate, is thought to be the most common [2].

The reported incidence is 0.02–0.05% and the condition is predominant among females at a ratio of 25:1 [8,9]. The fourth metatarsal is most frequently affected followed by the first and the fifth toes and 72% of these cases occur in both feet [10]. Accompanying hypoplasia of the phalanx, hypoplasia of the soft tissue, brachydactyly, brachymetacarpia, or osteochondromatosis are common [9]. Patients seeking treatment commonly have cosmetic concerns. However, many may also complain of pain in the forefoot or toes, callusities, or problems with shoe wear due to toe alignment and mechanical insufficiency of the short metatarsal [9,11,12].

Conservative treatment consists of accommodative shoe treatment and padding. This treatment regime is often unsatisfactory to the patient, because it does not alleviate the chief complaint of cosmesis [9]. Multiple surgical procedures have been described for brachymetatarsia. The most widely used ones include one-stage elongation with bone graft and gradual elongation by distraction osteogenesis with or without shortening of the adjacent metatarsals and phalanges, and shortening of adjacent metatarsals and phalanges [10,13–15]. Metatarsal lengthening for brachymetatarsia by a hydroxyapatite graft has also been reported [16].

The aim of this case series study was to evaluate the validity of gradual distraction of the fourth metatarsal to tackle this problem in adults.

2. Patients and methods

From March 2004 till April 2014, six patients of congenital shortening of the fourth metatarsal were referred to our center...
with bilateral brachymetatarsia in five cases [total: 11 feet]. Metatarsal shortening was congenital in all patients. All cases were females. Age of patients ranged from 19 to 30 years [average 23 years]. Shortening ranged from 8 to 24 mm [15–64% of the original length]. The second-fourth angle ranged from 38° to 47° with a mean of 41.7°. There was no functional problem preoperatively. Before surgery, we discussed the procedure in details with the patients. Hence, they became aware of the various complications associated with distraction osteogenesis (e.g., the temporary loss of motion and pin tract infection) and consented to distraction osteogenesis. Surgery aimed at anatomical restoration to recover proper weight bearing and to minimize cosmetic problems. However, the patients were concerned mainly by the cosmetic problem.

2.1. Operative technique

The patient was positioned supine on a radiolucent table, and a support was placed under the ipsilateral hip to internally rotate the leg and facilitate access to the lateral foot. Spinal or general anesthesia was used. There was no need for tourniquet use. The lengthening was performed using a rigid minifixator (Penning Fixator), and self-drilling, self-tapping screws with 2 mm diameter. This frame is unilateral with two proximal and two distal screws placed on the dorsolateral aspect into the proximal metaphysis and the distal metaphysis under fluoroscopic guidance. Sometimes, extra-pins were used. After the application of the screws, the frame was temporarily removed, and osteotomy was performed in the middle third through 1 cm incision (Fig. 1A). Multiple drilling by 1.6 mm K-wire was done before osteotomy. Then the frame was applied. Fluoroscopic imaging was used to check the completeness of osteotomy (Fig. 1B). Sterile gauze and crepe bandage were applied thereafter (Fig. 2A).

The patients were instructed to be non-weight bearing. After a waiting period of 12 days, distraction started at a rate of 0.75 mm/day. Then the rate was modified according to radiographic findings of callus formation and the degree of developing stiffness in the metatarsophalangeal joint. Radiographs were checked every other week to inspect the degree of osteogenesis and joint condition. Distraction was continued until a satisfactory metatarsal length had been achieved (Fig. 2B). After radiographic confirmation of consolidation, guarded weight bearing was initiated. The external fixators were removed when the callus had matured (Figs. 3 and 4). The frame was removed as an office procedure. Evaluation

![Fig. 1](image1.png)

*Fig. 1.* Intraoperative images. (A) Osteotomy was performed in the middle third through 1 cm incision. (B) Fluoroscopic image to show the completeness of osteotomy.

![Fig. 2](image2.png)

*Fig. 2.* Postoperative clinical photographs. (A) Postoperative clinical photograph showing brachymetatarsia of the 4th metatarsal with an external fixator applied. (B) Clinical photograph of bilateral fourth brachymetatarsia with right side corrected by external fixator.
depended upon the achievement of the target length, angulation, pain, and satisfaction of the patient.

3. Results

The outcome was assessed clinically according to the Japanese Society for Surgery of the Foot (JSSF) standard rating system of lesser metatarsophalangeal–interphalangeal length [17] and graded as excellent (>85), good (71–85), fair (56–70), or poor (<56). External fixation period was from the date of surgery to fixator removal. Healing index is calculated as the number of days of external fixation treatment per centimeter of distraction osteogenesis.

There were eight excellent and three good results after an average follow up of 2.6 years [range 6 months–6 years]. Average length gained was 15.4 mm [range 8–24 mm] and average length gained as a proportion of the original metatarsal length was 39.5% [range 15–64%]. The mean external fixation period was 69.1 days (63–84 days). The average healing index was 46 days/cm. Bony consolidation was achieved in all metatarsals. The mean JSSF score was 91 (85–100) points. After the observation period the gait was normal, no deformities were found in the metatarsals and no cases of residual pain were reported either. Patients reported esthetic and functional satisfaction with the results (Fig. 5).

Complications included mild pin tract infection in nine cases, which responded well to oral antibiotics. Subluxation of the metatarsophalangeal (MTP) joint developed in one case. Delayed regenerate formation occurred in two cases that were treated by repeated compression and distraction.

4. Discussion

We found that distraction osteogenesis effectively treated fourth brachymetatarsia with few adverse effects. Both cosmetic and functional results were good-to-excellent, and patient satisfaction was high. While many patients with brachymetatarsia seek treatment for cosmetic concerns, surgical reconstruction must also result in proper forefoot alignment and function. Restoring normal anatomy is important because brachymetatarsia may result in abnormal pressure distribution of the forefoot causing excessive load and pain at adjacent metatarsals [9,11,12]. Various surgical methods for the treatment of this malformation have been published; the main ones and the most widely used ones by several authors, with variable results, are: one-stage lengthening and bone grafting, and bone lengthening by callus distraction [13–15]. The advantages of one-stage metatarsal lengthening include a shorter period of bony union and better patient compliance [10].
However, this method has the disadvantages of donor site morbidity, neurovascular impairment due to rapid stretching, and a poor length gain with undercorrection due to the tension in the surrounding soft tissues [14,18,19]. In fact, neurovascular compromise is generally considered the major limitation to acute lengthening beyond 10 mm [20]. Baek and Chung [21], in a series of 34 one stage metatarsal lengthening, reported four cases of metatarsophalangeal stiffness, two fractures, one loss of fixation and one delayed union. Lee et al. [22] reported patient dissatisfaction in 11.1% with one-stage intercalary bone grafting. Some authors perform combined adjacent metatarsal or phalan- geal shortening to reduce the associated complications and the period of treatment [1,20]. A gradual intraoperative soft-tissue distraction technique, using a bone spreader, for 20–30 min could help to avoid critical neurovascular compromise. But excessive pressure of the spreader may cause fracture of the osteotomized bone end [1,20]. While one stage lengthening procedures such as osteotomy and interposition are more appropriate for correction of moderate length deficiencies, the callus distraction method is more suitable for larger elongation [23].

Congenital shortness of the metatarsal bone represents one of the best indications for distraction osteogenesis in the foot [15]. The advantages of distraction osteogenesis include: no need for bone grafting, easier tendon stretching, fewer neurovascular complications, early weight bearing, and a larger length gain, whereas disadvantages include stiffness or subluxation of the metatarsophalangeal joint, pin tract infection, and a longer period of bony union [10–13]. Of specific importance in metatarsal lengthening is the preoperative physical and psychological assessment and screening of patients in addition to the preoperative patient awareness of the details of the procedure and its possible complications. This is very important when considering cosmetic concerns and its psycho-logical impact particularly in female patients.

Frierson et al. [24] found no differences in the pattern of bone healing and the amount of newly formed bone after corticotomy or osteotomy and the use of the oscillating saw may lead to delayed consolidation. We performed the osteotomy with pre-drilling using 1.6 mm K-wire. Pre-drilling was done in order not to extend the osteotomy into the nearby half-pin. To minimize heat generation we used slow speed interrupted drilling with cooling by cold saline. Osteotomy after pre-drilling markedly improved the results of Lee et al. [22] compared to the use of microsaw. Lamm [20] used 1.8 mm K-wire for pre-drilling.

With distraction osteogenesis, some authors have reported healing indices of 60, 64, and 74.6 days/cm [1,10,12] but the healing index we obtained in our series was 46 days/cm. The reason behind that might lie in the fact that we used a small incision for the osteotomy, minimal dissection, and low energy corticotomy. According to Hwang et al. [25], the healing index was about 3.8 months/cm in callotasis for adults with first brachymetatarsia.

JSSF rating system [17] was chosen to clinically assess the outcome of our series because it is comprised of four site-specific scales. It has the lesser metatarsophalangeal–interphalangeal scale that is devised for disorders or trauma of the second to fifth metatarsals and their phalanges and joints. We considered this a more specific scale rather than a scale for the whole foot. In addition, it covers many aspects regarding pain, function (activity limitation, footwear requirements, MTP joint motion, IP joint motion, MTP-IP joint stability, and any painful callus or clavus), and alignment.

Loss of any motion or loss of more than 50% of dorsiflexion has been considered to be MTP joint stiffness in several studies [11,12]. Some authors recommend the fixation of the proximal phalanx using an accessory clamp distally or the introduction of a Kirschner wire to prevent iatrogenic subluxation of the MTP joint during distraction. However, erosion of the cartilage of the metatarsal head after temporary intramedullary fixation by K-wires and limitation of movement had been reported before [1,17,18]. In our series, joint stiffness was not a problem because we started active and passive MTP joint mobilization exercises from the first postoperative day. As the patients were aware of the possible complications, they were very cooperative regarding the regular active and passive exercises. The only patient who was not strictly following the instructions developed subluxation of the MTP joint and that was resolved after fixator removal and vigorous physiotherapy. Also, the MTP joint motion was checked regularly in the follow up visits and the rate of distraction was modified according to the degree of developing stiffness in the metatarso- phalangeal joint. Lee et al. [10] observed MTP joint stiffness in 13 of the 27 feet and Shim and Park [11] reported 23% MTP joint subluxation or stiffness.

Lee et al. [10] reported callus fracture in three feet and explained this by too rapid distraction rate and walking with weight bearing using the whole sole, which would result in overloading of the callus during ambulation. They also reported two pin breakages that occurred during the lengthening period in two patients. We did not encounter any fractures or pin breakage. This may be because guarded weight bearing is initiated only after radiographic confirmation of consolidation. Delayed regenerate formation occurred in two cases that were treated by repeated compression and distraction. This might be due to relatively rapid

Fig. 5. (A and B) Clinical photograph showing corrected right 4th brachymetatarsia with full range of motion.
distraction and/or some periosteal injury. Patients reported esthetic and functional satisfaction with the results. The scar of the procedure was not an obstacle for patients with bilateral brachymetatarsia to ask to operate the contralateral side after their satisfaction with the result of the operated side.

5. Conclusions

We recommend gradual lengthening of the 4th metatarsal as the treatment of choice for shortness of the 4th toe. Soft tissue release or fixation of the metatarsophalangeal joint to guard against displacement was not needed.

Conflict of interest

The authors declare that they have no conflict of interest.

References