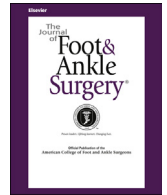




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Case Reports and Series

Mini-Invasive Treatment for Brachymetatarsia of the Fourth Ray in Females: Percutaneous Osteotomy With Mini-Burr and External Fixation—A Case Series

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ABSTRACT

Brachymetatarsia is a rare disease defined by metatarsal shortening and characterized by aesthetic dissatisfaction with or without pain. The aim of our study was to evaluate the outcomes of fourth ray brachymetatarsia treated with percutaneous osteotomy using a mini-burr and gradual lengthening with external fixation. A total of 7 females were recruited for the study; 6 (85.71%) of whom had a bilateral deformity, for a total of 13 feet affected by fourth ray brachymetatarsia. Percutaneous diaphysis osteotomy with a mini-burr followed by metatarsal elongation was performed. Metatarsal lengthening was measured as the difference between the preoperative and postoperative length at external fixator removal. The American Orthopaedic Foot and Ankle Society lesser toe metatarsophalangeal-interphalangeal score, patient satisfaction, restoration of Lelièvre parabola, and treatment time were evaluated. Numerical data are reported as the mean $\pm$ standard deviation and 95% confidence intervals. The Mann-Whitney U test was used to compare the changes in the AOFAS score with a level of significance of $p < .05$. The mean metatarsal lengthening was 17.46 ± 4.89 (95% confidence interval [CI] 14.8 to 20.12) mm and the mean treatment time was 99.23 ± 8.53 (95% CI 94.59 to 103.87) days. The mean American Orthopaedic Foot and Ankle Society lesser toe metatarsophalangeal-interphalangeal score improved significantly from 76.38 ± 2.66 (95% CI 74.77 to 78.03) preoperatively to 86.46 ± 1.45 (95% CI 84.85 to 88.07) postoperatively ($p < .01$). In 12 of 13 feet (92.31%), the Lelièvre parabola was restored, and the patients were satisfied with the clinical outcomes. The results of our study demonstrate that percutaneous osteotomy with the mini-burr and external fixation is an effective treatment for lengthening of fourth ray brachymetatarsia. Furthermore, we found good clinical and functional outcomes, high patient satisfaction, and a similar duration of treatment compared with other gradual lengthening procedures.

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Brachymetatarsia is a rare foot deformity caused by the premature closure of the metatarsal epiphysis that leads to a shortened metatarsus (Figs. 1 and 2). Several factors seem to play a role in the etiology of this foot condition, including traumatic, iatrogenic, and congenital, with or without an association with systemic syndromes (1). This deformity is characterized by a wide range of clinical patterns from aesthetic dissatisfaction to severe pain. By definition, it is the presence of ≥ 1 metatarsals ending 5 mm before

the parabolic metatarsal arc (2). It is more frequent in females (25:1), and the fourth ray is the most frequently affected (3), with an incidence of 0.02% to 0.05% (4). In 75% of cases, it presents as a bilateral condition (5).

Several procedures have been proposed for the surgical correction of this deformity to restore the Lelièvre parabola. Gradual lengthening by distraction elongation (6) and one-stage elongation seem to be the most commonly used procedures. One-stage elongation is usually combined with hydroxyapatite (7) or bone grafting (8) and, when necessary, is completed with an adjacent metatarsal and phalange shortening osteotomy (9).

In our retrospective case series, we describe the results of gradual metatarsal lengthening performed with mini-burr percutaneous osteotomy followed by external fixation.

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Fig. 1. Clinical photograph showing fourth ray brachymetatarsia.

Patients and Methods

From December 2012 to July 2015, we recruited 7 consecutive females with congenital metatarsal shortening of the fourth ray. Six of the patients (85.71%) were affected by bilateral brachymetatarsia, for a total of 13 rays (toe + metatarsal). The mean age was 25 (range 18 to 30) years. The amount of shortening was 11 to 24 mm. The inclusion and exclusion criteria are listed in Table 1. The demographic characteristics of the included patients are listed in Table 2. Before surgery, the patients were fully informed of the surgical procedure, treatment duration, and potential complications. All 7 patients provided written informed consent and agreed to undergo the procedure. The aim of surgery was to restore the normal anatomy and biomechanics of the metatarsal arc, thereby also improving the aesthetic aspect of the deformity. We measured the metatarsal lengthening after percutaneous surgery as the difference between the length before treatment and after external fixator removal. The Lelièvre parabola was considered to be restored within <5 mm of tolerance (10). The American Orthopaedic Foot and Ankle Society (AOFAS) lesser toe metatarsophalangeal-interphalangeal score (11,12), patient satisfaction, and period required for external fixation were also recorded.

Operative Procedure

The procedures were performed by the same surgeon (F.Z.), with the patient in the supine position and under spinal anesthesia. A tourniquet was not used. A support was placed under the ipsilateral hip to internally rotate the leg and facilitate access to the lateral foot. Lengthening was achieved using a minifixator (Minirail™, Orthofix, Verona, Italy) and self-drilling, self-tapping screws with a 2-mm diameter. The screws were positioned at the level of the proximal and distal metaphyses under fluoroscopic guidance. After screw



Fig. 2. Radiograph showing fourth ray brachymetatarsia.

Table 1

Overview of inclusion and exclusion criteria for patient selection

Inclusion criteria	
Congenital brachymetatarsia of fourth ray	
Age >18 yr	
Pain or aesthetic dissatisfaction	
Metatarsal ending ≥ 5 mm before parabolic arc	
Exclusion criteria	
Brachymetatarsia of a different ray	
Brachymetatarsia secondary to trauma or foot surgery	
Brachymetatarsia secondary to hereditary syndrome or associated with syndactyly and polydactyly	

placement, the frame was removed. A 2-mm skin incision was performed at the level of the middle third of the metatarsal diaphysis to insert the mini-burr (Shannon 44™, Miltex Instrument, Co., Inc., Riethem-Weilheim, Germany) for osteotomy. This procedure was completed under fluoroscopic guidance. Finally, the 2 metatarsal fragments were separated by about 3 mm, and a frame was applied. Sterile gauzes and bandages were used to cover the screw portals (Fig. 3). The first postoperative day, full weightbearing was allowed with an orthopedic shoe (Podalux™, DJO Global, Surrey, United Kingdom) without the use of a cane or other support.

Postoperative Care

The patients were instructed in the care of the external fixator and how to medicate the pin tracts (Fig. 4) and to begin the lengthening at a rate of 0.25 to 1 mm/day starting 4 to 5 days postoperatively. Radiographic evaluations were performed 20, 45, and 90 days postoperatively before external fixator removal. The patients underwent clinical examinations every 10 days and used a personal elongation rate related to radiographic callus formation and stiffness developing at the metatarsophalangeal joint. Distraction was continued until the necessary metatarsal length was reached. The external fixator was removed as an outpatient procedure (Fig. 5). Patients continued to wear the orthopedic shoe for the next 3 weeks.

Statistical Analysis

The numerical data are reported as the mean $\pm$ standard deviation and 95% confidence intervals (CIs). The Mann-Whitney *U* test was used to evaluate the changes in the AOFAS lesser toe metatarsophalangeal-interphalangeal score. The level of significance was set at $p \leq .05$.

Assessors

Two of us (F.F., F.L.) performed the radiographic measurements, data collection, outcome assessments, statistical analysis, and language editing. Two others (O.C., F.S.) recruited the patients and performed the

Table 2

Main demographic characteristics

Demographic Characteristic	Value
Participants (N)	7 (100)
Female gender (N)	7 (100)
Age (yr)	
Mean	25
Range	18 to 30
Laterality (n)	
Bilateral (12 rays in 6 patients)	6 (85.71)
Left (1 ray in 1 patient)	1 (14.28)
Patients with aesthetic dissatisfaction (N)	7 (100)

Data in parentheses are percentages.



Fig. 3. Clinical photograph showing external fixator in place.



Fig. 4. Clinical photograph showing medication of the pin tracts.



Fig. 5. Clinical photograph showing the final outcome.

radiographic measurements. Finally, 1 of us (F.Z.) performed the radiographic measurements, surgery, and outcome assessments.

Results

At external fixator removal, the mean metatarsal correction was 17.46 ± 4.89 (95% CI 14.8 to 20.12) mm. The mean duration for external fixator use was 99.23 ± 8.53 (95% CI 94.59 to 103.87) days (Table 3). The mean follow-up period was 14.77 (range 6 to 24) months. At the last follow-up visit, all the patients were satisfied with the procedure and the aesthetic and functional results.

The mean AOFAS lesser toe metatarsophalangeal-interphalangeal score improved significantly from 76.38 ± 2.66 (95% CI 74.77 to 78.03) before treatment to 86.46 ± 1.45 (95% CI 84.85 to 88.07) postoperatively ($p < .01$; Table 4).

In 12 of 13 feet (92.31%), the Lelièvre parabola arc was restored (Fig. 6). In 1 foot (7.69%), early consolidation arrested the elongation process 5 mm short of the desired length. One case (7.69%) of domestic trauma with pin rupture was surgically treated. In that case, a protracted elongation time of 20 days was needed to reach the required length.

Discussion

The surgical strategy for brachymetatarsia is still controversial, and the postoperative results have been jeopardized by the frequent complications. In particular, one-stage lengthening seems to be

Table 3

Mean metatarsal elongation at external fixator removal and mean time of external fixator use (N = 13 rays in 7 patients)

Outcome	Mean $\pm$ SD	95% CI
Elongation (mm)	17.46 ± 4.89	14.8 to 20.12
Operative time (min)	99.23 ± 8.53	94.59 to 103.87

Abbreviations: CI, confidence interval; SD, standard deviation.

Table 4

Mean American Orthopaedic Foot and Ankle Society lesser toe metatarsophalangeal-interphalangeal score before and after treatment (N = 13 rays in 7 patients)

Variable	AOFAS Score
Preoperatively	
Mean $\pm$ SD	76.38 $\pm$ 2.66
95% CI	74.77 to 78.03
Postoperatively	
Mean $\pm$ SD	86.46 $\pm$ 1.45
95% CI	84.85 to 88.07
p Value	<.01

Abbreviations: AOFAS, American Orthopaedic Foot and Ankle Society; CI, confidence interval; SD, standard deviation.

associated with graft reabsorption and the risk of aesthetic concerns such as donor site scarring and skin necrosis (5,8). External fixators, however, have shown a high risk of infection, misalignment, joint subluxation, and delayed consolidation (13).

Despite the wide range of possible complications, only 1 complication developed in our series, a case of pin rupture due to domestic trauma. No cases of delayed consolidation developed, although 1 case of early consolidation occurred. Early consolidation seems to be related to a low elongation rate or the use of an osteotomy procedure (14). In particular, the use of a mini-burr could be associated with an increase in bone particles in the osteotomy site. This event, according to Bastos Filho et al (15), can improve the regeneration rate and callus formation. However, burr rotation generates heat that could be a possible cause of bone sclerosis, with a risk of delayed consolidation. Therefore, during the surgical procedure, we irrigated the mini-burr blade with cold saline solution.

Weightbearing, as tolerated, was allowed from the day after surgery with appropriate shoe wear and without support, when possible.



Fig. 6. Postoperative radiograph showing restoration of the Lelièvre parabola arc.

Podalux™ (DJO Global) footwear has a low dynamic load on the metatarsal head (16), and the early ambulation stimulates edema reabsorption and bone healing owing to the resulting muscle contraction and mechanical load (17).

The small “tissue-sparing” incision (18) using a mini-burr and the bone and bone marrow particles produced during the osteotomy (19) seem to be key factors for good callus formation and reducing the risk of delayed consolidation. The early weightbearing also reduces joint stiffness and the length of the rehabilitation program, which for several of our patients was not necessary.

Owing to the rarity of this condition, little is known about the surgical strategy. Gradual lengthening seems to have the ability to permit greater correction compared with a one-stage procedure. Lee et al (20) compared 3 different surgical procedures (“one-stage lengthening”, “distraction osteogenesis using a saw,” and “distraction osteogenesis using osteotomy”) for brachymetatarsia in terms of the postoperative results according to the AOFAS score and metatarsal lengthening. Their study showed similar results in terms of the AOFAS score for all groups. However, the distraction osteotomies seemed to be associated with greater and more statistically significant amounts of correction (20).

Our series demonstrated results comparable to those of other research in terms of treatment time, amount of surgical correction, and clinical outcomes (20–23). However, the small sample size and the rupture of pins in 1 case, which required a protracted external fixator time, could have influenced the treatment time.

Some bias must be related. Some are intrinsic to the rarity of the pathologic entity itself and the study design, such as the small number of patients and the lack of a control group and randomization. Also, an a priori sample size calculation was not done. We were also limited by the lack of published data with which to compare our results. We believe more high-quality studies and longer follow-up periods are necessary to confirm our results and curtail some of the study bias.

In conclusion, despite the limits of our research, we can conclude that performing percutaneous osteotomy with a mini-burr and external fixation is an effective treatment strategy for lengthening fourth ray brachymetatarsia. The lack of the complications typical with external fixator use, delayed consolidation, and joint stiffness and the presence of good clinical outcomes and high patient satisfaction make it a valuable treatment choice.

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