

## Treatment for Brachymetatarsia of the Fourth Ray via Lengthening Over the Pin: A Case Series

### Abstract

**Background & Aim:** Brachymetatarsia is a rare condition of the foot characterized by a shortening of the metatarsal bones. Brachymetatarsia may be congenital and idiopathic in etiology, or caused by trauma, and may be associated with systemic diseases. This deformity is recognized with a wide range of clinical patterns, from aesthetic dissatisfaction to severe pain. Various surgical methods have been proposed to treat the disease, which could be associated with different complications such as stiffness, lack of union, and deviation from the axis. The present study aimed to evaluate the result of "over the pin" surgical method for patients with brachymetatarsia.

**Methods:** This case series research was performed in a training hospital on the patients referred to the orthopedic clinic, with congenital brachymetatarsia confirmed by foot radiographs. Data related to the demographic characteristics of patients, the obtaince length, and the duration between treatment onset and removal of the fixator were obtained using a checklist and the medical files of the participants. In this study, the exclusion criteria were non-metatarsal involvement, trauma, secondary foot surgeries, genetic syndromes, syndactyly, and polydactyly.

**Results:** There were women and one male, with the mean age of 12.3 years. Bilateral involvement was detected in two female subjects, and a total of 11 surgeries were assessed. According to the results, there was a total length increase of 16.58 millimeters over a mean duration of 96.5 days. In addition, metata length increased by 48.23% during treatment, and no malunion was observed in the patie

**Conclusion:** "Lengthening over a ping" for brachymetatarsia, of fourth ray is superior to other techniques, which avoids malunion, and higher length gain is possible.

to the results of the study, malunion was reported in none of the participants after the surgery, which showed an apparent superiority of the method, compared to other techniques. However, a higher length increase was detected in the current research, compared to other studies.

**Keywords:** Brachymetatarsia, Surgical Effectiveness, Ilizarov Apparatus

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### Introduction

Brachymetatarsia is a rare forefoot deformity caused by premature closure of the epiphyseal plate of the metatarsal, which leads to abnormal shortness of a metatarsal bone. Various factors such as trauma, iatrogenic and congenital issues are involved in the formation of this problem with and without association with systematic syndromes<sup>(1)</sup>. This deformity is recognized with a wide range of clinical patterns, from aesthetic dissatisfaction to severe pain. In general, brachymetatarsia is defined as more than one metatarsal ending five mm or more proximal to the parabolic arch of the other adjacent metatarsals. This condition is 25 times more prevalent in females compared to males<sup>(1)</sup>. In most cases, the fourth toe is affected by a general prevalence of 0.02% to 0.05%, and the disease emerges bilaterally in 75% of the cases<sup>(3)</sup>. Several surgical methods have been proposed to correct this syndrome, some of the most widely used of which include gradual metatarsal lengthening and the one-stage metatarsal lengthening. It is notable that the latter is often combined with hydroxyapatite and bone grafting<sup>(4, 5)</sup>.

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There are different types of gradual lengthening, such as callus destruction<sup>(6)</sup> and gradual metatarsal lengthening by external fixation through the skin<sup>(7)</sup>. In this respect, one of the treatments is lengthening over the pin for gradual metatarsal lengthening. Given the rarity of brachymetatarsia, no research has been conducted on the treatment of the disease in the country. However, some foreign studies have been performed in this area, the results of which are presented below:

- In a research, H. T. Kim et al. (2003) evaluated 18 patients treated with one-stage metatarsal lengthening along with a bone graft, and the results were indicative of a healing index of 1.3 months/cm and 30% increase in the length after the treatment<sup>(8)</sup>.
- In 1999, Masada et al. conducted a research on six patients using the callus destruction method. In the end, an improvement was observed in all patients and a mean length increase of 30% was observed. Moreover, the average lengthening index was 82 days/cm<sup>(9)</sup>.
- In a research by Fusini et al. (2017) on seven patients, who underwent gradual metatarsal lengthening by external fixation, the mean increase in length was 17.43 mm during the average time of 99.70 days<sup>(10)</sup>.

This case series study was conducted to evaluate the effectiveness of lengthening over the pin surgery on patients with brachymetatarsia.

## Method

This retrospective, case series research was performed in the healthcare training complex of Rasoul Akram Training Hospital in Iran University of Medical Sciences in 2018. The research population included patients with brachymetatarsia of the fourth ray referred to the mentioned complex during 2017-2018. Due to the rarity of this condition, all of the patients referred to the orthopedic clinic, whose congenital brachymetatarsia was confirmed by an orthopedic specialist based on foot radiographs, were enrolled in the study.

Data related to the demographic characteristics of patients, the increased length, and the duration between treatment onset and removal of the fixator were obtained using a checklist and the medical files of the participants. The exclusion criteria were non-metatarsal involvement, trauma, secondary foot surgeries, genetic syndromes, syndactyly, and polydactyly. Subjects were selected by the observational method and non-random convenience sampling. Given the nature of the research, all patients meeting the inclusion criteria were entered into the study. The results were presented in the form of mean $\pm$ SD and percentage for quantitative and qualitative variables, respectively. Moreover, data analysis was performed in SPSS version 20. The participants ensured of the confidentiality terms regarding their personal information, and informed consent was obtained from the patients prior to the study. It is notable that the research was confirmed by the ethics committee of the Iran University of Medical Sciences with an ethical code of IR.IUMS.FMD.REC 1396.8821215155, and the researchers adhered to the principles of the Declaration of Helsinki.

## Results

The research was performed on nine patients with brachymetatarsia, eight of whom were female and one was male with a mean age of  $12.3 \pm 2.87$  (min and max ages of six and 16 years, respectively). Since two female patients had bilateral brachymetatarsia, a total of 11 surgeries were evaluated. The information obtained from patients is shown in Table 1. According to the results, a mean length increase of  $16.58 \pm 4.89$  mm was observed in a mean period of  $96.5 \pm 8.53$  days. Moreover, a mean increase of  $48.23 \pm 10.11\%$  was observed in the metatarsal length of the participants during the treatment period. In addition, malunion was reported in none of the participants. Various treatment stages of five patients are presented in figures one-three.

**Table 1. Demographic characteristics, lengthening a treatment duration of patients**

Patient	Gender	Age	Lengthening level	Treatment period days
1	Female	14	20.8	96
2	Female	12	20.9	92
3	Female	6	10.8	89
4	Female	12	16.8	101
5	Female	15	20	100
6	Female	12	20.6	86
7	Female	10	15.1	103
8	Female	10	12.4	81
9	Male	16	21.6	91

## Discussion

This case series study was performed on nine patients with brachymetatarsia treated with lengthening over the pin surgical method, who

were followed up for consecutive time intervals. The prevalence of the disease is more than 25 times higher in females compared to males, which is consistent with our findings. In the first session of treatment, the Ilizarov apparatus and a pin were planted in the metatarsal (with a two-week interval). The pin remained in patients' body until reaching an acceptable length and was removed in the last session with radiographic control. Due to the longitudinal pinning, no deviation was observed in growth. Moreover, there was no report of secondary infection and joint problems. the primary treatment for patients with brachymetatarsia is non-surgical measures such as medical insoles, orthotics, and wide toe box orthopedic shoes. Surgery is indicated if surgical procedures could not reduce the patient's pain and there were aesthetic incentives<sup>(4)</sup>.



**Figure 1. Patient number two**

There are still doubts about the surgical treatment of brachymetatarsia due to the numerous and sometimes dangerous post-operative involvements. The amount of bone lengthening is an important issue to consider before the surgery, which is measured by various formulas. In addition, several multiple-stage metatarsal lengthening surgical techniques have been described for correcting the problem<sup>(11)</sup>, one of the most common of which are lengthening by bone grafts and sequential increases by callotaxis via an external fixator<sup>(12)</sup>. In a one-stage metatarsal lengthening surgical technique, there are concerns about scars after tissue grafting, skin necrosis, and adsorption of grafted tissue<sup>(13)</sup>. In addition, gradual lengthening surgery increases the risk of tissue infection, joint dislocation, lack of growth in the desired direction, and delayed ossification<sup>(2)</sup>.

In a review study, Jones et al. (2015) evaluated various treatments of brachymetatarsia and

compared the one-stage bone grafting with callus destruction and a combined technique. Among the three methods, most complications (both major and minor) were related to the callus destruction method (complications were observed in 52% of patients) and the combined method, whereas the lowest level of complications was observed in patients treated with the one-stage metatarsal lengthening technique (19% patients<sup>(12)</sup>). Moreover, the most prevalent complications were metatarsophalangeal joint (MTPJ) and partial metatarsal dislocation. A solution for this problem is placing half an additional pin in the proximal phalanx with a short metatarsal bone<sup>(11)</sup>. Despite these complications, the callus destruction method had relative superiority over other methods in the present study owing to a higher bone lengthening in the patients treated with this technique.



Figure 2. Patient number three





Figure 3. Patient number four

Despite the wide range of expected complications, no growth delay was observed in this case series. On the other hand, premature bone growth was found in one case, which might be due to a low increase in length or osteotomy with a pin. In a research, Fuiano et al. (2019) used callus destruction with an external fixator to treat 12 patient with brachymetatarsia for a mean duration of 22 months. The average increase in length was 16.8 mm (with a range of 8-22 mm). The complications and morbidity of this procedure were low (in four surgeries), which included infection, one case of medial deviation of the metatarsal bone, and two brief MTPJs. In addition, it was recommended that special attention be paid to patients with more than 20 mm shortness. It is notable that our findings are congruent with the results obtained by the aforementioned research<sup>(14)</sup>. In a retrospective research in 2009, researchers compared three different surgical methods to treat brachymetatarsia of the fourth ray. One group of patients was treated

with one-stage intercalary bone grafting supported by Kirschner Wire Implant Module. The second group was treated with gradual lengthening via an external mini-fixator after an osteotomy blade saw, whereas the third group received treatment by an external mini-fixator after osteotomy using pre-drilled holes. At the end, the results of 11%, 13%, and 0% of participants respectively in the first, second and third groups were unsatisfactory. Therefore, re-surgery was performed on 20 feet due to complications such as metatarsophalangeal Joint problems (seven cases in all groups), bone formation failure (four cases in the saw group), skin maceration (four cases in the bone-graft group), deviation from the axis (four cases in the bone-graft and saw groups) and external fixation fracture (one case in the saw group). None of these complications were observed in the present research with the exception of skin scars. Similarly, the mentioned complications were not reported by Fuiano et al. (2019)<sup>(15)</sup>. In another case report in 2013, a 16-year-old

patient with brachymetatarsia underwent treatment with lengthening scarf osteotomy held with an Omnitech. This technique allows a lengthening stage through a single incision with graft incorporation in six weeks<sup>(16)</sup>. According to the results, the application of a pin could lead to an increase in bone fragments at the osteotomy site, which, based on the results obtained by Bastos Filho et al.<sup>(17)</sup>, could improve ossification and faster callus formation. Patients were allowed to walk on their foot for a few days after surgery, which greatly affected the reduction of tissue edema. In the research by Fusini et al. (2017), surgery by diaphysis percutaneous osteotomy by mini-bar and external fixator was performed on 13 feet with brachymetatarsia. In line with our findings, the mean increase in the length was 17.46 mm. Moreover, the method was reported to have a similar recovery period, compared to multistage surgeries, and patients were adequately satisfied with the results<sup>(10)</sup>. Given the rare nature of the disease, there is an information gap about the suitability of the mentioned surgical methods. Our findings confirmed the results of other studies regarding the duration of treatment

and the amount of bone lengthening. However, a higher bone lengthening was detected in the current research, which showed that performing the surgery at younger ages is more beneficial to the person. Another cause of the superiority of the technique was using a pin that eliminated the possibility of unnatural growth. Given pinning in the longitudinal direction of the fourth metatarsal in patients, there was no deviation from the axis. It is recommended that future studies be conducted to evaluate various factors affecting the failure of the technique.

## Conclusion

Despite the limitations of the research, the method of lengthening over the pin was recognized as an effective surgical method for the treatment of the fourth metatarsal deformity. Other complications such as delayed growth and joint stiffness were not observed, and the complications observed (e.g., limited skin lesions) were acceptable. Therefore, the lengthening over the pin method could be used as an alternative valuable and effective technique.

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