

## CASE REPORT

## Bilateral Brachymetatarsia: A Rare Case Report

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### ABSTRAK

*Brachymetatarsia adalah suatu keadaan tidak normal pada bahagian tulang jari kaki metatarsal yang menjadi pendek kerana penutupan pramatang pada pertumbuhan plat epiphyseal. Keadaan ini selalunya terjadi pada bahagian tulang jari kaki metatarsal yang pertama dan tulang jari kaki metatarsal yang keempat. Punca keadaan ini boleh disebabkan oleh masalah kongenital atau disebabkan punca-punca yang lain. Kami ingin melaporkan kes seorang wanita berusia 53 tahun yang mengadu kesakitan kaki dan mempunyai masalah ketika ingin memakai kasut. Pemeriksaan pada bahagian kaki menunjukkan kedua-dua belah jari kaki keempat dalam keadaan pendek dan jari-jari kaki itu condong ke arah atas. Dia telah didiagnosis sebagai bilateral brachymetatarsia dan pembedahan telah dilakukan atas alasan kosmetik. Kemudian, proses pemanjangan tulang jari kaki metatarsal telah berlaku secara beransur-ansur dengan menggunakan external fixator MiniRail (Orthofix) dan K-wiring pada bahagian sendi metatarsal jari kaki keempat. Proses pemanjangan tulang jari kaki metatarsal keempat dan penyatuan tulang dapat dicapai dengan baik di akhir proses rawatan.*

*Kata kunci: kongenital, kesakitan, pemanjangan tulang*

### ABSTRACT

Brachymetatarsia is a conditioned which has been described as abnormal short metatarsal bone due to premature closure of the epiphyseal growth plate. It usually involves a single metatarsal bone with the fourth metatarsal being the most common. It may be congenital or acquired. We here report a case in a 53-year-old lady who presented with foot pain and shoe wearing problem. Examination revealed shortening of the bilateral fourth toe and the toes were tilted dorsally. She was diagnosed with bilateral brachymetatarsia and operation was done mainly for cosmetic reasons. Gradual lengthening with distraction osteogenesis was

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performed using a MiniRail external fixator (Orthofix) and K-wiring of the right fourth metatarsophalangeal joint. Adequate length of fourth metatarsal and good bony union were achieved at the end of treatment.

Keywords: deformity, pain, lengthening

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## INTRODUCTION

Brachymetatarsia is an unusual diagnosis with an incidence of 0.02% to 0.05% (Shim & Park 2006). It predominantly affects females (F: M=98:4) and 72% of cases are bilateral (Kim et al. 2004). It can present in either unilateral or bilateral, in single or multiple metatarsal (brachymetapody) and in combination with syndactyly or polydactyly. In extremely rare case, the combination of bilateral brachymetatarsia and brachymetacarpia can occur together in individual patient (Aski et al. 2017). The causes include congenital, post-surgical, post-traumatic, and can be associated with diseases such as sickle cell anaemia, multiple epiphyseal dysplasia, juvenile rheumatoid arthritis, Apert's syndrome, Down's syndrome, Albright's osteodystrophy, and poliomyelitis (Baek & Chung 1998; Lamm 2010; Lee et al. 2010)

Patients with brachymetatarsia have high-riding toe on the dorsum of foot which causes difficulty in wearing shoes. It is often complicated with dorsal digital corns and painful plantar metatarsal head callus. They can also have short phalanges, toe dysplasia, transfer metatarsalgia, toe deformities and cosmetic reasons (Lee et al. 2010). In long standing brachymetatarsia, global transverse digital deviation

results in windswept deformity and bunion deformity formation.

The aim of surgery is to improve shoe wearing, restore weight bearing alignment, reduce the pain associated with the deformity and to achieve better cosmesis (Shim & Park 2006; Lamm 2010). The most widely used techniques for lengthening are either acute, single-stage lengthening with interposition bone graft or gradual distraction by callotasis with or without shortening of the adjacent metatarsal and phalanges (Baek & Chung 1998; Kim et al. 2003; Lamm 2010).

We report a case of bilateral brachymetatarsia treated with gradual lengthening by callotasis. The patient had low self-esteem and problems in shoe wearing due to her deformity.

## CASE REPORT

A 53-year-old female noticed that her bilateral fourth toes were short compared to other toes at the age of six years. There was no history of trauma during childhood. She complained of pain in both foot after prolonged walking and had difficulty to choose a proper foot wear. She also had problem of hygiene at the recessed fourth web, bilaterally. This deformity had affected her emotionally and her self-esteem. On examination, the fourth toes were shortened bilaterally and were tilted



Figure 1: Clinical photograph of bilateral short fourth metatarsals. L: left; R: right; arrows showing affected fourth metatarsals.



Figure 2: Anteroposterior radiograph of bilateral short fourth metatarsals. Arrows showing lytic lesion at both metatarsal heads.

dorsally (Figure 1). No skin ulcer or callosity was noted. The difference in length between the tips of fourth toe to the third toe was 1.5 cm. Her gait was normal and she was able to stand on toe tip and heels.

The radiograph of both feet showed lytic lesion of both metatarsal head (Figure 2). CT scan of both feet revealed hypoplastic fourth metatarsal and proximal phalanges bilaterally with normal joints. Tc99m MDP Whole Body Bone Scan and SPECT-CT were done and no abnormal findings observed. We proceeded with MiniRail external fixator (Orthofix) for lengthening of the right fourth metatarsal. Incision made over dorsum of right foot in between fourth and fifth metatarsal bone. Intraoperatively, we noted that the bone quality was soft. Under fluoroscopic guidance, three threaded wire size 2 mm inserted at proximal metaphysis of fourth metatarsal at 45° from the frontal plane and right angles to the

bone axis. MiniRail lengthener with clamps applied parallel to the plantar surface of foot. Another two threaded wire inserted at distal metaphysis. Osteotomy was performed with saw in between the two clamps. A K-wire inserted longitudinally through the fourth metatarso-phalangeal joint to prevent any subluxation during callus distraction (Figure 3).

After seven days post operation, 0.25 mm of distraction was applied two times daily (0.5 mm per day). The patient was seen in clinic once or two weekly and clinical and radiographic findings were assessed. The lengthening was continued until eight weeks post operation, the optimum length and satisfactory parabola of all five metatarsal heads was achieved. The external fixator was maintained in position until consolidation occurs. She was admitted at five-month post operation for removal of external fixator. Intraoperative, the third proximal pin broken and was

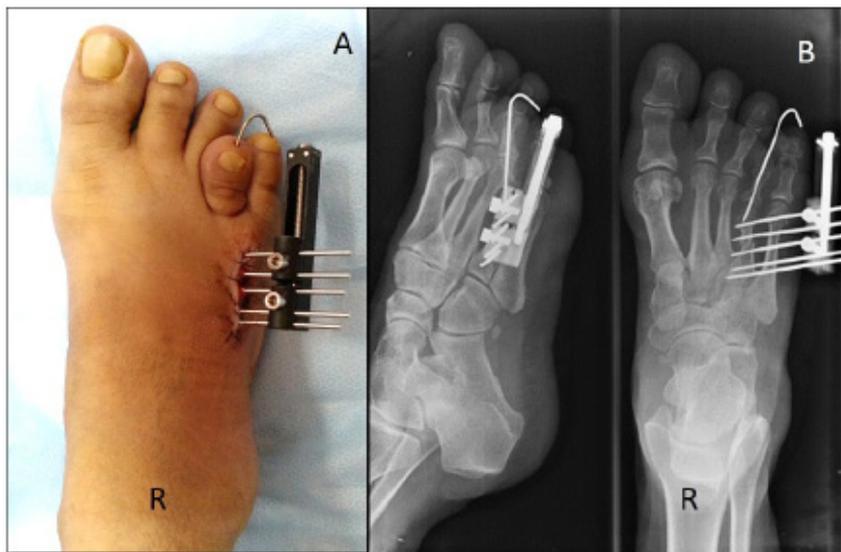


Figure 3: Immediate postoperative clinical picture (a) and radiograph (b) showing MiniRail external fixator and K-wire over fourth metatarsophalangeal joint. The osteotomy site was at midshaft of right fourth metatarsal. R=right

unable to be removed. Incomplete union of the distraction bone to the base of the phalanx was noted and two axial K-wires inserted to facilitate stabilization and hence union. Back slab applied and K-wire was removed after one month. Radiograph showed union of the distraction bone and clinically adequate length of fourth metatarsal bone achieved (Figure 4). She was able to ambulate well post removal of K-wire and was satisfied with the results. She was planned for lengthening of the left fourth metatarsal using the same method of external fixator.

## DISCUSSION

Patients with brachymetatarsia have difficulty in wearing shoes and are often complicated with painful plantar callus and dorsal digital corns. Operative treatment aims to correct the deformity

by lengthening the short metatarsal. One-stage lengthening procedures are preferred when the target length was less than 15 mm, whereas gradual lengthening is preferred for target length more than 15 mm (Kim et al. 2004; Lee et al. 2010).

McGlamry and Cooper were the first researchers to describe acute lengthening with autogenous bone graft in 1969 (McGlamry & Cooper 1969). Gradual distraction is applied intraoperatively with creep and stress relaxation of 20 to 30 mins duration. This technique allows shorter period for bony union with less scar formation (Baek & Chung 1998; Choi et al. 1999). Previous study reported success in single-stage lengthening with intercalary autogenous bone graft and application of gradual distraction during operation (Baek & Chung 1998). They concluded that gradual distraction technique intra-



Figure 4: Clinical photograph (a) showing correction of the fourth digits and proper length restoration. The skin was hyperpigmented and scar formation. Radiograph (b) showing complete union after nine month post operation. Note the broken pin at the proximal part of fourth metatarsal. R=right.

operative is effective, elementary and can overcome the deficiency of single-stage lengthening such as small gain in length and neurovascular damage (Baek & Chung 1998).

Gradual lengthening with distraction osteogenesis using an external fixator is preferred for lengthening of more than 1cm. Ilizarov semi-circular fixator, mini-Hoffmann and monolateral fixators are the options of external fixators used for this technique. The method of osteotomy is either open or percutaneous osteotomy. Percutaneous osteotomy is preferred because it limits devascularisation of surrounding tissues by preserving the endosteum and periosteum (Lee et al. 2010). This will reduce scar formation and improves new bone formation. Study of couple of year before concluded that gradual lengthening with osteotomy using an osteotome through pre-drilled holes

produced better results compared to gradual lengthening with osteotomy using saw and one-stage lengthening with interposition bone grafting (Lee et al. 2009).

The advantages of gradual lengthening includes the risk of neurovascular compromise is reduced, patient is allowed to fully weight bear during treatment, and the final postoperative length can be predicted and achieved without secondary soft tissue lengthening or additional bone graft (Shim & Park 2006; Lamm 2010; Lee et al. 2010). The attachment of external fixator for a long duration is very uncomfortable especially for young adult and adolescent. It can result in scar formation at the pin sites, temporary hyperpigmentation of skin around pin sites and pin track infection (Kim et al. 2004). Stabilization of MTPJ by Kirschner-wire (K-wire) pinning

of the metatarsal head to the digit reduces the risk of MTPJ subluxation during the lengthening period. Other complications include over-lengthening, transverse and sagittal plane malalignment and longer time for bony union (Kim et al. 2004; Lamm 2010).

In lengthening surgery for brachymetatarsia, preoperative determination of amount of lengthening is required to re-establish the metatarsal parabola (Lamm 2010). Based on the length required the time of distraction and consolidation can be estimated. During postoperative period, the desired rate of distraction 0.5 mm per day may need to be adjusted according to clinical and radiographic assessment during follow-up to avoid under-lengthening or over-lengthening. The consolidation period typically ranges from 14 to 18 weeks but varies depending on the location of osteotomy, patient's age, comorbidities, medications, smoking status, amount and rate of metatarsal lengthening (Lamm 2010).

This patient was complicated with poor healing and delayed union of the distracted bone to the distal osteotomy site. The younger patient has a better healing capacity and they recommended that older patient who has lower healing capacity need to be observed and follow-up, closely (Shim & Park 2006). The aim of surgery was achieved as patient had satisfactory length of toe, enhance shoe wearing and better foot appearance.

## CONCLUSION

Brachymetatarsia can be corrected by various surgical methods with promising outcomes in terms of cosmesis, improve shoe wearing, functional and anatomical restoration and patient satisfaction. The choice of treatment depends on severity of foot deformity, age of patient, surgeon and patient preferences.

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