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## Case Report

# A rare case of isolated congenital unilateral brachymetatarsia presenting in a young adult male

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## ABSTRACT

Congenital brachymetatarsia is a rare skeletal anomaly which is characterized by the abnormal length reduction of one or more metatarsal bones. It occurs as the result of the early closure of the growth plate. While the majority of the reported cases are idiopathic, it can also be seen in association with various genetic conditions or syndromes. Brachymetatarsia most commonly involves the fourth metatarsal followed by the first metatarsal. This rare disorder is mostly bilateral and reported almost exclusively in females. Herein, we present a rare case of congenital unilateral brachymetatarsia which was incidentally detected in a young adult male.

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## Introduction

Brachymetatarsia refers to an abnormal reduction in the length of one or more toes. The disorder may be congenital or occur secondary to trauma or surgery. Congenital brachymetatarsia is a rare skeletal anomaly of which the incidence ranges between 0.02% and 0.05%. It occurs as the result of early closure of the growth plate and mostly involves the fourth metatarsal followed by the first metatarsal. In rare cases where more than one metatarsal of the same foot is involved, the most common combination is the first and fourth metatarsals. While the majority of the reported cases are idiopathic, congenital brachymetatarsia can also be seen in association with various genetic conditions or syndromes [1]. Trisomy 21, monosomy X, pseudohypoparathyroidism,

Al-bright syndrome and diastrophic dysplasia are disorders related to congenital brachymetatarsia [2].

In general, the development of the anomaly begins during the embryonic period and continues as the child grows, causing deceleration of the entire morphogenetic process of the metatarsal. The anomaly can be detected during childhood when the physes are still open. However, it usually becomes evident before the definite closure of the growth plate of the metatarsal, which corresponds to the age of about 14 [3]. While there is not yet a well-established objective criteria for the determination of brachymetatarsia, Martinez et al proposed a new diagnostic test that enables quantification of the shortening of the fourth metatarsal in brachymetatarsia. Using this method, they aimed to determine the metatarsodigital alterations most frequently related to this deformity [4].

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**Fig. 1 – Clinical photograph of both feet of the patient showing that the fourth finger of his left foot is shorter than its counterpart.**

Congenital brachymetatarsia is reported to affect almost exclusively females (96% of the cases) and mostly show bilateral involvement (72% of the cases) [5]. Herein, we present a rare case of congenital unilateral brachymetatarsia which was incidentally detected in a young adult male.

### Case report

A 17-year-old male who was a candidate for army officer admitted to our hospital for mandatory health screening. He did not have any health complaints. He is the second of 3 sons born to nonconsanguineous healthy parents. There is no family history of any kind of congenital skeletal abnormalities in his immediate or extended family.

On physical examination, the fourth finger of his left foot was shorter than its counterpart (Fig. 1). On anteroposterior radiograph of both feet demonstrated that the fourth metatarsal was shorter than normal. The rest of the right metatarsals, all left metatarsals, all phalanges, and tarsals of both feet were regular in shape and number. Lateral and lateral oblique radiographs of the right foot showed no abnormality other than the shortness of the fourth metatarsal (Figs. 2 and 3).

Comprehensive evaluation of the patient, including detailed physical examination, laboratory tests, thoracoabdominal radiographs, and abdominal ultrasonography examination within the scope of compulsory health screening, did not reveal any other pathologic finding. Based on the characteristic radiographic findings, the patient was diagnosed as having isolated unilateral brachymetatarsia. Since the patient had no health or cosmetic complaints, no treatment was planned. However, albeit minor, this skeletal anomaly was a condition disqualifying for the officer candidate.



**Fig. 2 – Anteroposterior radiograph of both feet of the patient demonstrating that the fourth metatarsal is shorter than normal. Note the rest of the right metatarsals, all left metatarsals, all phalanges, and tarsals of both feet are regular in shape and number.**



**Fig. 3 – Lateral (a) and lateral oblique (b) radiographs of the right foot showing that the fourth metatarsal is shorter than normal. Note the rest of the bone structures and all articular relations of the foot are normal.**

## Discussion

The clinical presentation of brachymetatarsia may vary depending on factors such as age, sex, number of the affected metatarsal, and uni/bilaterality of the involvement [1]. Patients with brachymetatarsia mostly apply for health care during adolescence or young adulthood. Since the exact closure of the growth plate of metatarsals occurs at about 14 years of age which corresponds to adolescence, the disorder presents with a complaint of cosmetic disturbance intensifying the body image anxiety, a characteristic feature of adolescence. This is not because the disorder is not visible at an earlier age, but because it begins to be perceived as a problem during this life period by the patient. The deformity turns out to be esthetically unacceptable for most of the affected subjects, and this is especially true for female adolescents. On the other hand, adults with brachymetatarsia most commonly present with pain (transfer metatarsalgia), irritation of the corresponding skin by the shoe wear or difficulty in walking [6,7]. The patient we currently present did not have any slight cosmetic disturbance or complaint of pain. Moreover, he was surprised to see that, unlike the other candidates, we obtained extra radiographs and photographs of his foot. Our case raises the question that brachymetatarsia could be more prevalent in the general population than previously reported. Many people with this minor anomaly, especially males, may not care at all and may not consult a doctor. And, given the known difference between the intensity of concerns about the body image between the male and female adolescents, the established acceptance that brachymetatarsia almost exclusively involves females may be incorrect.

Since our patient was asymptomatic, no treatment was planned. In symptomatic patients, management of the deformity may be either conservative or surgical. Various surgical techniques have been developed for the purposes of achieving an aesthetically acceptable foot and relieving metatarsalgia. The most commonly used surgical techniques for the correction of brachymetatarsia are elongation with bone graft interposition and sequential lengthening using callotasis by means of external fixators [8]. Recently, Fusini et al have reported a retrospective case series of brachymetatarsia of the fourth metatarsal which they performed gradual metatarsal lengthening with mini-burr percutaneous osteotomy followed by external fixation. They achieved good clinical and functional results with high patient satisfaction over a similar treatment duration compared to other gradual lengthening techniques. And they recommended this method for the treatment of brachymetatarsia involving the fourth metatarsal [9]. For the management of brachymetatarsia involving the first metatarsal, Kim et al reported that performing one-stage step-cut lengthening of the metatarsal combined with shortening and/or lengthening of the adjacent metatarsal and phalangeal bones, they obtained excellent cosmetic and functional results [10].

In conclusion, congenital brachymetatarsia is a rare minor skeletal anomaly which presents in adolescence or young adulthood with cosmetic discomfort and, in some cases, with pain. By applying current surgical procedures, both cosmetic

problem and pain can be resolved successfully. Our case suggests that the actual information regarding the frequency and gender preference of this deformity may be questioned.

## Informed consent

Informed consent for publication was obtained from the patient.

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## Declaration of Competing Interest

The authors declare that they have no conflict of interest.

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