Os Intermetatarseum Revisited: A Case Report of Rare Variant and Review of Literature

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ABSTRACT

Os intermetatarseum is the rarest accessory bone of the foot. It is usually found between 1st and the 2nd metatarsal bases arising typically from the base of the 2nd metatarsal. Only a few symptomatic cases have been reported in the literature, which were either unilateral or bilateral and radiographically they were of different shapes and sizes. We present a large, bilaterally symmetrical, fully formed variant of os intermetatarseum, fusing to both metatarsals has not been described before. The case report also describes the surgical anatomy during the excision of os intermetatarseum and review of the literature to date.

Keywords: Os intermetatarseum, Accessory bone of the foot, Metatarsal bar, Deep peroneal nerve compression.

CASE REPORT

A 14-year-old boy, who does karate at professional level, presented with a 4 months history of pain and prominence on the dorsum of both feet. Pain aggravates during his karate training and especially when he sits on his heels. He was also complaining of intermittent pins and needles radiating to the 1st and 2nd toes.

On examination, he had normal gait, had prominence on the dorsum of both feet (Fig. 1). Both feet have normal medial arch and overall alignment of both lower limbs is normal. Palpating over the prominence caused mild discomfort and was bony hard in consistency. There were no neurovascular deficits, but Tinel’s sign was positive on percussing over the base of the swelling. There was no hypermobility of the hallux. He had full range of movement in his ankle, subtalar, midfoot and forefoot. Knees, hips and spine were clinically normal. He had no generalized joint laxity.

Radiographs were obtained which demonstrated bony mass bridging between the first and second metatarsal (Fig. 2). With a differential diagnosis of osteochondroma a further CT scan was performed which clearly demonstrated the fused separate ossification center at the base of the second metatarsal and fully formed os intermetatarseum fused to the first metatarsal (Fig. 3).

INTRODUCTION

Gruber first described os intermetatarseum way back in 1877. Ever since, only a few cases of symptomatic os intermetatarseum have been reported. The incidence of os intermetatarseum quoted in the literature from cadaveric and radiological studies ranges from 0.2 to 12.5%. Coskun et al2 identified 0.2% incidence in a radiographic study among 984 Turkish patients. Cilli found an incidence of 1.2% in 464 radiographs. Tsuruta et al4 studied the accessory bones of foot and ankle in 3460 radiographs of patients over a period of 7 years and observed that os Intermetatarseum is the rarest of the accessory bones of the foot and ankle with an incidence of 2.6%. Pfitzer5 documented a 12.5% incidence in his report of 520 cadaver dissections. Os intermetatarseum is usually bilateral and arises most commonly from the base of the 2nd metatarsal. It can be rudimentary or a fully formed bone and can be of any shape or size.6,7 In our case, os intermetatarseum represents a large bony bridge between 1st and 2nd metatarsal.

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Fig. 1: Prominence over the dorsum of both feet
Fig. 2: Os intermetatarseum bridging 1st and 2nd metatarsal

Fig. 3: Computed tomography reconstruction showing os intermetatarsaum with ossification center at the base of the 2nd metatarsal

Fig. 4: Neurovascular bundle underneath the EHB

After consultation with the parents and with the patient, excision of os intermetatarseum of both feet was performed.

Surgical technique itself is an exercise in dissecting the neurovascular bundle and protecting them throughout the procedure (Figs 4 to 6).

Interoperatively, on both sides we noticed identical findings of inflammation and thickening of the deep peroneal nerve, just above the large os intermetatarsaum. Os intermetatarsaum itself was found arising from the base of the second metatarsal and extending distally to fuse with the shaft of the first metatarsal distally. This was excised and it measured $5 \times 1 \times 0.5$ cm on both sides (Fig. 6). At 3 months follow-up he was symptom-free and was allowed to go back to sports. At the final follow-up of 18 months he remained symptom-free and was back to his professional karate and plays all other sports without any problems. Radiographs at the final follow-up were normal with no further regrowth (Fig. 7).

DISCUSSION

Os intermetatarsaum is a rare but well-documented accessory bone of the foot. It could be an incidental finding on routine radiographs or diagnosed when they become symptomatic. It can present as unilateral, bilateral, typically arising from the base of the 2nd or 1st metatarsals but can very rarely arise between 4th and 5th metatarsals. Various theories have been put forward. It is commonly believed that it is a form of polydactylysm. Henderson in his article proposed that this bony spur as the lost 1st plantar interosseous muscle and as a contributing factor for the development of the hallux valgus deformity in a case series of four patients. Friedl believed that it is a sesamoid bone of the first dorsal interosseous muscle from the calcification of the accessory tendon of the dorsal interosseous muscle. Our case demonstrates a complete bar extending from the base of the second metatarsal and fusing to the 1st metatarsal symmetrically in both feet suggestive of probable polydactylysm or poly metatarsal etiology.

We did an extensive literature search and found that, from the time it was first described in 1877, only 16 symptomatic cases of os intermetatarsaum were reported. Reichmister presented three cases in a 19, 25 and 44 years old individuals who presented with dorsal foot pain. Knackfuss et al reported a case of os intermetatarsaum, compressing on the medial branch of the deep peroneal nerve in a 52-year-old female patient. Smith and Welch reported a case of painful
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It appears that symptomatic os intermetatarseum is usually presented during preadolescent or adolescent age groups and mainly symptomatic with sport related activities. Nakasa et al13 from Japan described four cases in athletes. Ruffing et al14 from Germany described this entity in two athletes. Noguchi et al15 reported a painful os intermetatarseum in a soccer player. Waters16 in 1958 and Scarlet et al17 in 1978 reported a case of painful os intermetatarseum. In all these cases, symptoms were consistent with dorsal foot pain or compression neuropathy of the deep peroneal nerve. However, there is evidence-suggesting association between os intermetatarseum, metatarsus primus varus and hallux valgus.6,18 Henderson6 in his series of 4 patients described metatarsus primus varus and hallux valgus due to os intermetatarseum. Noguchi et al18 reported a case of bilateral hallux valgus associated with os intermetatarseum. Our patient was a 14-year-old karate player with deep peroneal nerve symptoms due to the stretch and compression of the nerve over the os intermetatarseum. In all these cases, symptoms improved after the excision of os intermetatarseum.

Compression of the nerve occurs typically between the os intermetatarseum and Extensor Hallucis Brevis (EHB). EHB forms the key anatomical landmark during the excision of os intermetatarseum. Deep peroneal nerve and dorsalis pedis artery lie just underneath EHB (Fig. 4) and can safely be protected by retracting EHB subperiosteally. One needs to be extremely careful in excising the proximal end of the os intermetatarseum, as the branch of the dorsalis pedis artery can be just coursing in close proximity between the base of 1st and 2nd metatarsal to form the deep plantar arch (Figs 5 and 6).

The deep peroneal nerve branches into medial and lateral branches just distal to the ankle mortise. Lateral branch provides motor innervation to extensor digitorum brevis (EDB) and EHB. Medial branch of the deep Peroneal nerve, courses over the talonavicular joint capsule and lies lateral to the 1st tarsometatarsal joint, then passes deep to the EHB tendon, where it is susceptible for compression between the EHB and os intermetatarseum. It further continues forward and bifurcates just before it terminates to give sensory innervation to the first web space.

**CONCLUSION**

Os intermetatarseum is a rear accessory bone of the foot. Young athletes presenting with dorsal foot pain and symptoms of deep peroneal nerve compression should raise the suspicion of this entity and should be a part of differential diagnosis of the dorsal foot pain. It is best investigated with radiographs and a CT scan. Magnetic resonance imaging scan may be needed rarely if not identified on a radiograph.19 Os intermetatarseum can be treated successfully by surgical excision.

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**Fig. 5:** Os intermetatarseum, between first and the second metatarsal. Arrow showing dorsalis pedis artery in close proximity to os intermetatarseum, coursing in between the base of 1st and 2nd metatarsal.

**Fig. 6:** Excision of os intermetatarseum. Arrow showing dorsalis pedis artery.

**Fig. 7:** Postoperative radiograph of both feet at final follow-up.
REFERENCES


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