Distal metatarsal coalition: A rare case report

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ABSTRACT

INTRODUCTION: Metatarsal coalition is an extremely rare condition. We report the second documented case of 4th and 5th distal metatarsal coalition in the literature.

PRESENTATION OF CASE: An eight-year-old girl was referred to an orthopaedic clinic with a four-month history of forefoot pain and swelling on the plantar aspect of the right little toe. Radiograph and clinical examination confirmed distal metatarsal coalition between the 4th and 5th metatarsals. Following a period of conservative treatment, excision was eventually performed due to worsening symptoms. Patient re-attended two years later with a recurrence of the coalition confirmed by computed tomography (CT) scan. The case was discussed at a tertiary paediatric orthopaedic institution. Decision was made to manage patient conservatively with insole and physiotherapy until skeletal maturity. A year later, patient’s symptoms did not worsen, and her foot displayed no evidence of change in the arch and shape.

DISCUSSION: The timing of ossification of coalition varies from one anatomical site to another. Surgery when performed before ossification is complete runs the risk of recurrence.

CONCLUSION: Our case report illustrates the importance of restoring normal weight bearing dynamics and pain relief when managing metatarsal coalition, or synostosis in skeletally immature patients. We recommend persevering with conservative treatment, with operative treatment reserved only as a later option, and ideally, until skeletal maturity is achieved.

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1. Introduction

Coalition, or synostosis, occurs when two adjacent bones are abnormally bridged by an osseous bar. Coalition of the foot is an uncommon abnormality and usually involves the hind-foot and mid-foot. Tarsal coalition is the commonest, majority of which affects the calcaneonavicular and talocalcaneal joints [1]. Metatarsal coalition remains extremely rare [1–6], with our case being the second documented case of coalition occurring at the level of distal metatarsal.

2. Presentation of case

An eight-year-old girl presented with a four-month history of forefoot pain and swelling on the plantar aspect of the right little toe. This had resulted in a significant pain and discomfort on walking; and had especially interfered with her sporting activities. There was no previous history of trauma, infection or any other joint problem. She had a normal birth history and achieved all developmental milestones appropriately. There was no family history of any hereditary syndromes associated with abnormality of the extremities.

Clinical examination revealed a prominence of the 5th metatarsal head with callosity developing over the plantar aspect. This was more pronounced on standing. Ankle, subtalar and metatarsophalangeal joint motions were normal. Radiograph of the foot demonstrated coalition between the fourth and fifth metatarsals, characterised by the metatarsal bar between the two metatarsals and bowing of the 4th towards the 5th metatarsal (Fig. 1). The physeal plate of the 5th metatarsal was also found to be pointing laterally (Fig. 1).

The parents were advised that operation could be an option but there is a risk of recurrence. The recommendation given to the parents was to continue with conservative management until skeletal maturity. However as the patient became more symptomatic, a decision was made to excise it. Microscopic analysis demonstrated a mixture of osseous, cartilaginous and fibrous tissue. The histopathology report concluded this to be a combination of osseo-fibrous-cartilaginous coalition. The patient continued to make good progress and was asymptomatic at four months following operation. She was later discharged from follow up.

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At age 10, the patient represented with swelling at the base of her right 5th metatarsal. Both patient and parents denied any injury or trauma since her initial surgery. The base of right 5th metatarsal was painful on palpation. There was also a reduced range of motion of the 5th metatarsal in relation to the 4th metatarsal. Repeat X-ray and CT scan with three dimensional reconstruction images confirmed a recurrence of the distal metatarsal coalition between the 4th and 5th metatarsals (Figs. 2 and 3).

The case was further discussed at a tertiary paediatric orthopaedic institution. A decision was made to manage patient conservatively with insole and physiotherapy until skeletal maturity. The patient is currently being followed up on an annual basis. During her most recent follow-up at age 11, patient’s symptoms did not worsen, and her foot displayed no evidence of change in the arch and shape.

3. Discussion

Metatarsal coalition, or synostosis, is uncommon relative to tarsal coalition. There are several ways to classify coalition. Perman and Wertheimer [7] classified coalition as congenital or acquired.

Most reported cases are congenital coalition between the bases of fourth and fifth [1–3]; or first and second metatarsal bones [4,5]. With only one other case in the literature [6], our case adds to the literature the second case of 4th and 5th congenital distal metatarsal coalition.

Histologically, coalition can be classified into one of the four types: fibrous, cartilaginous, osseous, or as illustrated by our case, a combination of all three tissue-types [8]. Based on symptomology, coalition can be classified as asymptomatic or symptomatic [9]. It has been reported in adolescents where ossification is incomplete, that asymptomatic cases can become symptomatic when exposed to risk factors such as increased activities [10], abnormal pedal biomechanics [11], locomotion on uneven terrain, ankle strain or sprain [12,13], and increased body weight [14]. It is possible that at age 8, increased sporting activities resulted in the development of symptoms in our young active patient where ossification is still not complete.

Coalition is also known to be associated with hereditary syndromes, such as Pfeiffer–Kapferrer syndrome [15], Cenani–Lenz syndactyly [16,17], Townes–Brocks syndrome [18], and Aperts syndrome [19]. At age 8 and with no family history, prior injury, trauma or infection, our patient has a symptomatic, non-syndromic congenital metatarsal coalition.

The timing of ossification of coalition varies from one anatomical site to another. Ossification happens in talonavicular joints at 3–5 years, in calcaneonavicular joint at 8–12 years, and talocalcaneal joints at about 12–16 years [20]. In our patient, histopathology of the excised sample taken at 8 years of age demonstrated presence of all three tissue-types (fibrous, cartilaginous and osseous). It is therefore likely that surgery occurred before complete ossification of the synostosis has occurred, and hence the recurrence following operative treatment.

Conclusion

Our case report illustrates the importance of restoring normal weight bearing dynamics and pain relief when managing metatarsal coalition in skeletally immature patients. The risk of recurrence with operative treatment should be explained to both patient and parents. In the presence of symptoms, we recommend persevering with conservative treatment. Operative treatment should only be reserved as a later option, and ideally, until skeletal maturity is achieved.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy
of the written consent is available for review by the Editor-in-Chief of this journal on request.

**Conflict of interest**

The authors declare that there is no conflict of interests regarding the publication of this paper.

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