Case Report

A rare case of bilateral congenital metatarsal synostosis

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We report a rare case of bilateral congenital metatarsal synostosis of the first and second metatarsal bones in a 6-year-old female, who was initially referred with obscure swellings of the soles of the forefeet (at the heads of the 2nd metatarsals) and intermittent pain on weight-bearing. Clinical examination had raised suspicion of a bony connection between the first and second metatarsals. This was confirmed by radiographs. Surgical intervention in the form of bilateral osteotomies proved successful in establishing pain free and more natural weight-bearing. © 2001 Harcourt Publishers Ltd

Introduction

Synostosis, a condition where two adjacent bones are abnormally bridged by an osseous bar, may be either congenital or secondary to injury (Atar et al. 1992). Examples include cranial synostosis, sacral synostosis, radio-ulnar synostosis (Giffet et al. 1986), navicular–cuneiform synostosis, metacarpal synostosis and metatarsal synostosis. Of these, metatarsal synostosis (MTS) is distinctly rare, only few cases having been reported in the world literature (Boccio et al. 1984, Kashuk et al. 1991, Keenleyside & Mann 1991a, b). These have all involved the 4th and 5th metatarsals.

We present what is, to our knowledge, the first case of synostosis between the first and second metatarsals. The first recorded case of MTS actually predates much of orthopaedic surgical history. This condition was radiographically confirmed in an archaeological specimen of an Eskimo in conjunction with unilateral ectrodactyly and hypoplasia of the foot (Keenleyside & Mann 1991a, b).

More recently, cases of synostosis of the foot have been reported, primarily involving the midfoot and hindfoot. Less commonly, the metatarsals have been implicated, similar to cases of metacarpal synostoses described (Buck-Gramcko & Wood 1993, Kawabata et al. 1997).

Case report

A 6-year-old female was referred to the Orthopaedic Out-patient Clinic with a history, since birth, of swellings on the plantar aspect of both forefeet, specifically in the region of the second metatarsal head. These swellings had been increasing in size as the child grew and were diagnosed on separate occasions as bony exostoses and bone cysts by general practitioners. Specialist intervention was prompted when the girl complained of pain after long periods of weight-bearing.

Examination of this patient revealed several unusual features. The most striking was that, unlike normal feet, where the ball of the great toe (first metatarsal head), was a primary area of weight-bearing in the forefoot, this had been replaced by the second metatarsal head, with the great toe held clearly off the ground.

Palpation revealed obvious immobility of the 1st and 2nd metatarsals relative to each other and loss of the transverse arches. A bony

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fusion of the first and second metatarsals was clinically suggested. Radiographs confirmed this diagnosis, clearly showing radio-opaque bars between the two bones and with no distinct margins (Figs 1 & 2).

Surgical intervention was decided upon, the goal being to separate the affected metatarsals, hence restoring normal configuration and function of the feet. Through curved, longitudinal incisions on the dorsum of each foot, the osseous bars were exposed and osteotomies performed. After surgery, bilateral bootee casts were used to immobilize the metatarsals, with the 1st being in plantar flexion.

Weight-bearing commenced 2 weeks postoperatively and at 4 weeks both casts were removed. Full, pain-free weight-bearing was achieved after 6 weeks. The second metatarsal head remained weight-bearing during the toe-off phase of the gait. This remained unchanged at 6 months and 1 year follow-up.

Discussion

This case highlights a diagnostic dilemma, especially since pathology of the foot such as this is often masked until true functional disability occurs. In addition, metatarsal synostosis is a rarity, which would be suspected by few clinicians. Further, this is a previously unreported site of synostosis. It is not surprising, therefore, that the initial diagnosis entertained by the primary care physicians, was that of a bilateral bone cyst of the second metatarsal heads. Our examination revealed that what had been described as a lump was actually the second metatarsal head, the heavily callused surface being a testament to its increased function of weight-bearing.

The consideration of this being an atavistic feature was doubtful in view of the primate foot exhibiting a large space between the first and second toes as well as clawing rather than fusion, the former sometimes seen in trisomy 21 (Wilkins 1994, Prasher et al. 1995). Also, no such abnormality has ever been classified with other atavistic malformations (Egawa 1977, Verhulst 1996).

Fortunately, this case was detected at an early age and had a favourable functional if not anatomical outcome. In comparison, the recognized clinical entity of synostosis of the 4th and 5th metatarsal is a functionally less severe a condition but it generally presents at a later age and is more difficult to correct as patients already may have irreversible changes at diagnosis. Although we were unable to correct the position of the metatarsal heads relative to each other, the patient's painful weight-bearing still resolved, possibly because the heads were now mobile rather than fixed. We believe that she is at risk of recurrence of this symptom if she undergoes significant weight-gain, or attempts high-impact sports and even the natural rigours of adulthood and ageing and would need long-term follow-up.

Pain-free weight-bearing is the major aim in treating metatarsal synostosis, whereas achieving

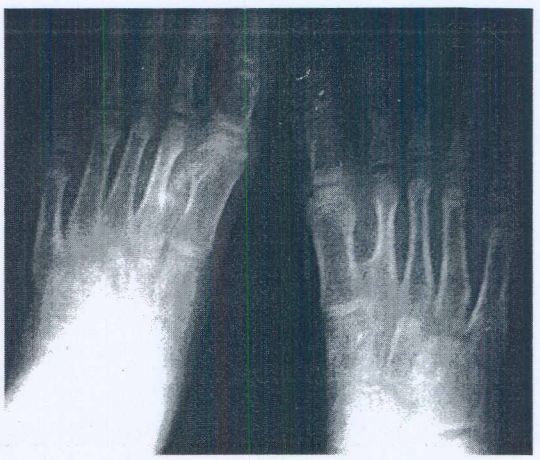

Fig. 1 Antero-posterior radiograph of left and right feet showing osseous bars. The left extends from the medial proximal end of the 2nd metatarsal as a straight bar to the lateral distal end of the 1st metatarsal. The right exists as a bony web at the proximal angle of the 1st and 2nd metatarsals with its distal edge concave.

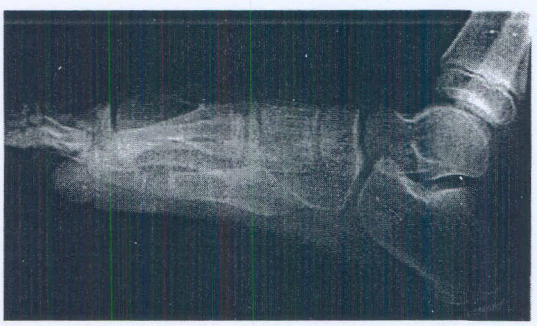

Fig. 2 Lateral radiograph of the right foot with the 1st metatarsal axis in a plane superior to that of the 2nd metatarsal which projects obliquely and inferiorly as it approaches the metatarso-phalangeal joint.

finer, more precise movements would be an essential goal in correcting metacarpal synostosis (Miura 1988, Ueba & Seto 1997), another documented condition whose treatment mode was considered in the context of our case. Thus, osteotomy, immobilization in a functional position and physiotherapy are adequate for metatarsal synostosis. In retrospect, though bony release was achieved, the end result might have been improved by metatarsal osteotomy and internal fixation to allow the 1st metatarsal head its intended role of weight-bearing, the second head to be elevated and the arch of the foot restored.

Conclusion

Although metatarsal synostois is a rare condition, early diagnosis, corrective surgery and physiotherapy can provide excellent results. Because the primary operative goal is to achieve pain-free weight-bearing, surgery is less technically demanding than the correction of metacarpal synostosis, where fine precise movements are an important aspect of hand function. Metatarsal osteotomy plus internal fixation to restore normal anatomy may provide satisfactory results.

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