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Bilateral Triple Talocalcaneal, Calcaneonavicular, and Talonavicular Tarsal Coalition: A Case Report

Ajmal Farid, MD¹, Frank W.M. Faber, MD, PhD²¹ Resident, Department of Orthopaedic Surgery, Haga Hospital, The Hague, The Netherlands² Orthopedic Surgeon, Department of Orthopaedic Surgery, Bergman Clinics, Rijswijk, The Netherlands

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ABSTRACT

We report a rare case of a female, aged 42 years, with symptomatic bilateral triple tarsal coalition, that is, talocalcaneal, calcaneonavicular, and talonavicular tarsal coalition. The patient was treated conservatively by adjusting her activities. At the 12-month follow-up, the patient was asymptomatic. Bilateral triple tarsal coalition is a rare disorder, especially in nonsyndromic patients. The purpose of this case report was to highlight this rare type of multiple bilateral tarsal coalitions and to discuss the relevant existing literature.

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Tarsal coalition is an abnormal bony (synostosis), cartilaginous (synchondrosis), or fibrous (syndesmosis) union between 2 bones of the hindfoot and the midfoot. The overall incidence of tarsal coalition range from 1% to 2% (1–4). The most common tarsal coalitions are calcaneonavicular (53%) and talocalcaneal (37%), which are seen in 90% of all cases (4). Other tarsal coalition such as talonavicular, calcaneocuboid, and cubonavicular are less common and represent 10% or less of the overall incidence of 1% to 2%. Bilateral tarsal coalition occurs in 50% of all talocalcaneal coalitions and in 60% of all calcaneonavicular coalitions (5) (Table). The prevalence of bilateral multiple tarsal coalition is less well-documented in the literature.

In this case report, we present a rare case of a female, aged 42 years, with bilateral symptomatic talocalcaneal, calcaneonavicular, and talonavicular tarsal coalitions without congenital anomalies or syndromes. We also discuss a summary of the relevant existing literature.

Case Report

A female, aged 42 years, woman presented to our outpatient clinic in January 2017 with history of bilateral progressive foot and ankle pain in both feet. Her pain increased with sports activities such as jogging and decreased with rest. The left foot was more symptomatic than the right foot. There was no history of trauma and she had no known family

history of foot deformity or syndromes in the patient's siblings, parents, or children.

Physical examination showed rigid pes planovalgus (Fig. 1) and restricted range of motion in the hindfoot; mobility of both ankle joints was normal.

Weightbearing radiographs of both feet showed a small talar beak sign and the C sign (Fig. 2).

Computed tomographic scanning revealed bony coalition of the talonavicular (Fig. 3), calcaneonavicular (Fig. 4), and talocalcaneal (Fig. 5.) joints bilaterally. It also showed arthritic changes in the posterior facet of the subtalar joint in both feet (Fig. 6.), which is likely the cause of her pain. The patient was treated by adjusting her activities. At the 12-month follow-up, the patient was asymptomatic.

Discussion

The clinical manifestation of a coalition in the adult age mostly includes a history of ankle sprains owing to increased laxity of the lateral ligament (6) as the result of decreased subtalar motion. The abnormal bony, cartilaginous, or fibrous union may cause pain symptoms owing to degenerative changes of the joint, and in some cases owing to microfracturing or a stress fracture of the bony coalition (7). Furthermore, tarsal coalition is usually associated with fixed flat foot deformity sometimes with peroneal spasm (6).

Although bilateral single tarsal coalitions are quite common, bilateral multiple tarsal coalitions are rare and less well-described in the literature. In 1960, Kendrick (8) was the first to report 2 cases of dual bilateral calcaneonavicular coalitions and talonavicular coalitions. Since then, there

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Address correspondence to: Ajmal Farid, MD, Department of Orthopaedic Surgery, Bergman Clinics, laan van oversteen 20, 2289 CX, Rijswijk, The Netherlands.

E-mail address: aj.farid01@gmail.com (A. Farid).

Table
The most common tarsal coalitions

Type	Total percentage	Percentage bilateral
Calcaneonavicular	53%	50%
Talocalcaneal	37%	60%
Talonavicular, calcaneocuboid and cubonavicular	10%	unknown



Fig. 1. Hindfoot view demonstrating bilateral pes planovalgus deformity.

have been a few more case reports describing bilateral multiple tarsal coalitions in the literature (1,5,9,10). Triple or multiple bilateral tarsal coalition are even more rare, except when associated with a syndrome or conditions such as fibular hemimelus, Nievegert-Pearlman syndrome, Alpert syndrome, multiple synostosis syndrome, or hereditary symphalangism (2,11,12). Only a few cases, in nonsyndromic patients, of triple or multiple bilateral tarsal coalition have been described in the literature



Fig. 2. Weightbearing lateral view of ankle demonstrating bilateral C-sign and talar beak sign.

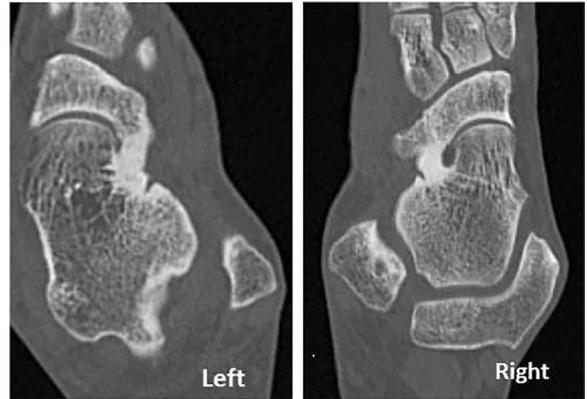


Fig. 3. Transversal computed tomographic scan demonstrating bilateral talonavicular coalition.

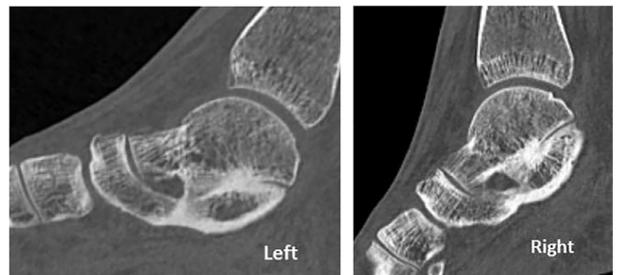


Fig. 4. Sagittal computed tomographic scan demonstrating bilateral calcaneonavicular coalition.



Fig. 5. Coronal computed tomographic demonstrating bilateral talocalcaneal coalition.

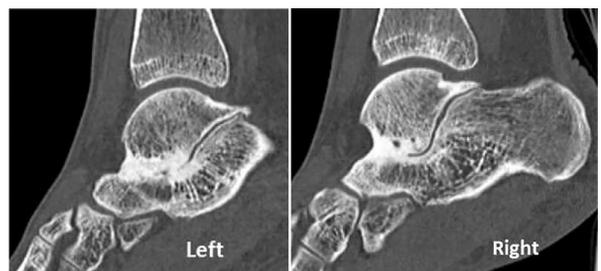


Fig. 6. Sagittal computed tomographic demonstrating bilateral arthritic changes in the posterior facet of the subtalar joint.

(13–15). The patient in our case report has no known condition or syndrome, as described. In her family, no syndromes or foot complaints have been documented and her children (2 females, ages 10 and 12 years) have completely normal feet without complaints.

In 2008, Goldcher (15) reported a case, similar to ours, of a female, aged 52 years, with bilateral triple talocalcaneal, calcaneonavicular, and talonavicular tarsal synostoses. The patient was successfully treated conservatively with orthosis. Subsequently, in 2013 Mettu et al (14) described a case report of a male, aged 35 years, with bilateral massive bony tarsal coalition involving the talus, calcaneus, cuboid, cuneiforms, and metatarsals. The patient was asymptomatic until the age of 35 years, whereafter he developed symptoms of secondary ankle arthritis, probably owing to the excessive stress of weightbearing activities. He was also successfully treated conservatively with rest, nonsteroidal anti-inflammatory drugs, and physical therapy (14).

The treatment of multiple tarsal coalition in adults is focused on symptom relief. Nonoperative management is the first choice and it includes physical therapy, orthoses, nonsteroidal anti-inflammatory drugs, physical activity restriction, and short leg cast immobilization. Surgical management of the tarsal coalition can be considered in patients with chronic symptoms and in patients with recurrent symptoms where the response to conservative treatment fails (16). Computed tomographic scans or magnetic resonance imaging is usually necessary to achieve the correct diagnosis. It is of essential value in the preoperative evaluation of the location and size of the coalition, and to evaluate if there is evidence for secondary adjacent joint arthritis (16). In our patient, isolated subtalar fusion could be considered when symptoms recur.

Triple bilateral tarsal coalition in nonsyndromic patients is very rare. Physicians must be aware of possible familial predilection. The treatment of triple tarsal coalitions should be focused of relief of symptoms. Operative treatment is only considered if conservative measures fail.

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