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Case Report

TIBIO-TALAR COALITION - A CASE REPORT AND REVIEW OF THE LITERATURE

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ABSTRACT

Background: A tarsal coalition is a bridging of two or more of the bones in the hindfoot. Multiple coalitions are described in the literature. However, the presence of a tibio-talar coalition has not previously been reported.

Results : Here, we describe the first reported case of tibio-talar and fibulo-talar coalition in the literature in a 31-year-old male. The patient had a 5-year history of bilateral hindfoot and ankle pain, with an established right sided talo-calaneal coalition. Investigations and subsequent open chilectomy and debridement surgery confirmed a tibio-talar and fibulo-talar coalition. The patient was discharged at one-year following surgery as he was pain free and returned to work.

Conclusions: We report the first case of failure of mesenchymal segmentation leading to tibio-talar fibrous coalition and fibulo-talar coalition. In this case surgical debridement provided resolution of symptoms. **Level of Clinical Evidence:** 4

KEY WORDS: Coalition, foot deformity, tarsal coalition, synostosis, surgery.

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INTRODUCTION

A tarsal coalition is a bridging of two or more bones of the hindfoot occurring as a result of failure of mesenchymal segmentation in utero. Although tarsal coalition affects 1-2% of the population [1,2], the true incidence may be higher as it is estimated that only 25% of patients become symptomatic [3]. Cadaveric and radiographic studies estimate the true incidence as between 6 and 13% [4–6].

An autosomal dominance method of inheritance with variable penetrance has previously been proposed [7,8]. Wray and Herndon described hereditary transmission of calcaneal-navicular coalition through three generations supporting this hypothesis [9]. There is no typical history that characterizes tarsal coalition but patients often describe a combination of the insidious onset pain with associated hindfoot stiffness and an inability to tolerate uneven walking surfaces. Examination often reveals little or no joint motion across the bridged segments of the foot.

Approximately, 90% of tarsal coalitions occur between the anterior process of the calcaneus and the plantar lateral aspect of the navicular (calcaneo-navicular) and the medial aspect of

the talus and the sustentaculum tali (talo-calcaneal) [2]. Other documented tarsal coalitions include talo-navicular, calcaneo-cuboid or naviculo-cuneiform joints but no previous accounts detail a failure of mesenchymal segmentation across the ankle joint [3].

Multiple coalitions have been described in the same foot of non-syndromic patients [10,11]. In almost 60% of non-syndromic patients with proven tarsal coalitions, abnormalities have been observed in both feet [2,4,12–14]. Tarsal coalition can occur as an isolated abnormality or in association with other syndromes. Coalitions are observed as part of fibular hemimelia, tarsal-carpal coalition, craniosynostosis, Apert syndrome and Nievergelt-Pearlman syndrome [13].

Radiographs of the feet should be obtained in assessment of any patient with suspected coalition. CT and MRI are useful in the further assessment of the coalition and surgical planning.

In this report, we describe a case with a known bony talo-calcaneal coalition on one foot and on the other, fibrous tibio-talar and fibulo-talar coalitions. We believe this to be the first case to be described in the literature.

CASE REPORT

A 31-year-old fit and well male of Pakistani origin presented with a 5-year history of worsening pain and stiffness in both ankles. At the time of presentation, his right side was more symptomatic of the two feet. On the right side, he localized pain and tenderness to the talo-navicular joint and it was noted that he had restricted ability to varise his hindfoot when standing on tiptoes. Ankle range of motion was normal.

On the left side, questioning revealed the pain and stiffness to be akin to classic anterior ankle impingement symptoms. There was no history of trauma nor of infection, inflammatory arthropathy nor any problems affecting gait in childhood. There was no history of consanguinity. There were no clinical features of fibular hemimelia, carpal coalition, craniosynostosis or Apert syndrome. Conservative measures including orthotics and analgesia offered no relief. On initial examination of his left foot and ankle, the patient demonstrated a symmetrical gait. He had some mild ankle swelling. His ankle range of motion was reduced with no dorsiflexion and 10 degrees of plantar flexion. He was tender along the anterior ankle joint line exhibited signs of anterior ankle impingement.

Imaging: Computer tomography (CT) images were obtained using a General Electric LightSpeed VCT 64 slice CT scanner and presented as multiplanar images (MPR) using a bone algorithm. Magnetic resonance imaging (MRI) was performed on a Philips Achieva 3T scanner using T1 turbo spine echo, T2 SPAIR (fat saturation) and proton density SPAIR (fat saturation).

Right side: CT and MRI scans confirmed synostosis between the sustentaculum tali and the middle facet of the talus on the right foot (Figure 1). Secondary features included marginal osteophyte (Figure 1d) and remodeling of the talo-navicular joint indicating chronic degenerative changes as a result of loss of supination and pronation in the subtalar joint. MRI confirmed stress changes and loss of joint space at the talo-navicular joint with dorsal and medial osteophytes. The ankle and calcaneo-cuboid joints were entirely normal as were the posterior and anterior facets of the subtalar joint.

Left side: On imaging of the left ankle, a CT and MRI demonstrated a fibrous coalition between the tibia and the talus (Figure 2,3,4,5) with exostoses arising from the lateral aspect of the neck of the talus and the anteromedial aspect of the distal tibial plafond. A fibrous fibulo-talar coalition was also noted (Figure 6). The other tarsal joints appeared healthy.

Surgical management:

The patient underwent staged procedures to both sides. As the right foot was more symptomatic, a talo-navicular fusion was performed in keeping with the clinical and radiological findings. Intra-operatively a degenerate joint was observed with very abnormal and unstable articular cartilage. The patient proceeded to arthrodesis and made an uneventful recovery from this procedure with complete resolution of pain.

A year after this surgery, the patient underwent

surgery on the contralateral ankle in the form of an open cheilectomy and debridement of the left ankle. Arthroscopic surgery had been excluded as an option because of the appreciable extent of the exostoses impeding adequate surgical access. Intraoperatively, the appearances of the exostoses suggested a congenital anomaly, akin to a failure of differentiation of the ankle joint. Of the two large bony exostoses, one projected from the anteromedial aspect of the distal tibial metaphysis towards the talus. The other arose from the anterolateral talus and projected towards the tibial metaphysis. Between the two was a sagittally-aligned cleft where the two exostoses almost interlocked.

Fig. 1: Right ankle. Bony coalition between the sustenaculum tali and the middle facet of the talus. Arrows pointing to coalition.
Top: MRI with coronal proton density, with fat saturation image a) coronal and b) sagittal.
Bottom: CT MPR c) coronal and d) sagittal MPR CT also showing bony coalition (arrow) and talo-navicular marginal osteophyte (circled)

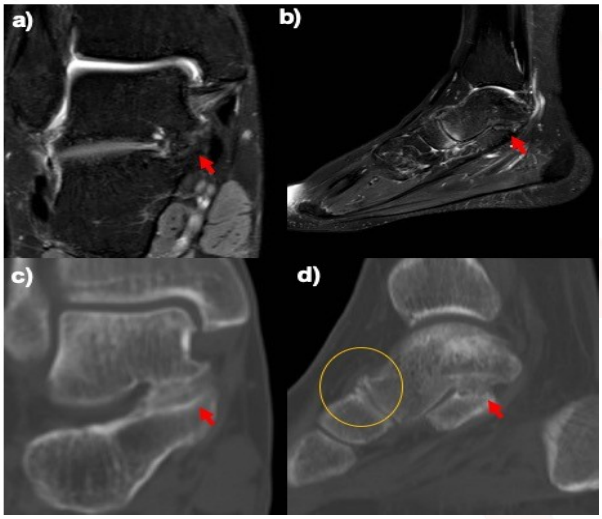


Fig. 2: Left ankle. MRI – a) sagittal T1 and b) CT - sagittal MPR showing an anterior, partly exostotic and partly fibrous, tibio-talar coalition. Arrows pointing to coalition.

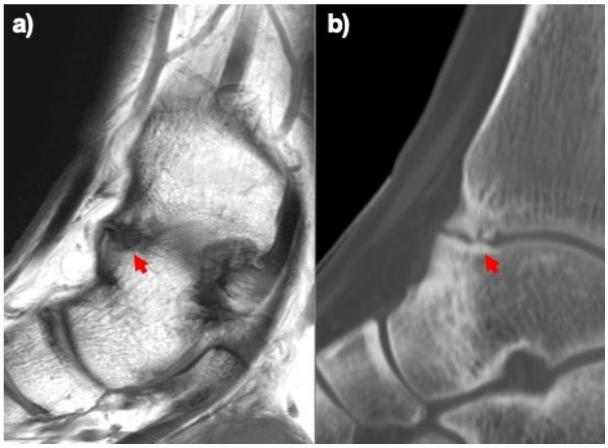


Fig. 3: a) Sagittal T1 and b) coronal proton density with fat saturation, showing an anterior exostotic and fibrous tibio-talar coalition. Arrows pointing to coalition.

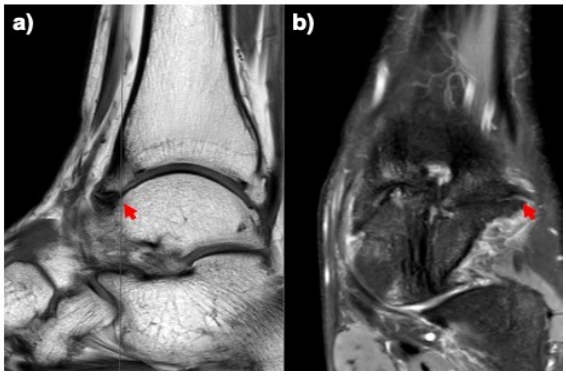


Fig. 4: Fibrous tibio-talar coalition – a) anterolateral and b) anterior ankle - CT Coronal MPR.

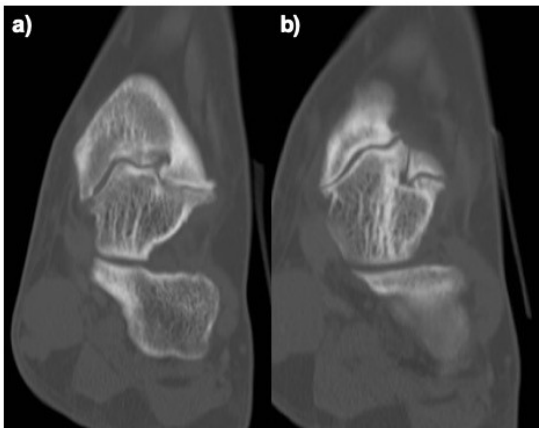


Fig. 5: 3D CT modelling of the right ankle joint showing the coalition.

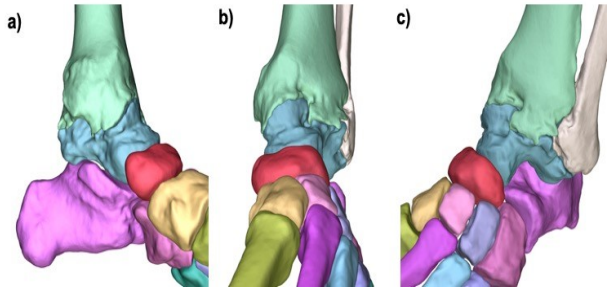
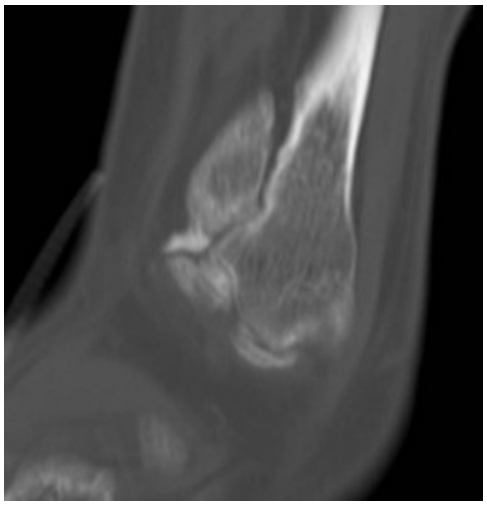


Fig. 6: Fibrous fibular-talar coalition CT sagittal MPR.



Post resection of the exostoses with osteotomes, they had the appearances of normal bone rather than osteophytes. The underlying joint surfaces were in good health. No attempt was made to release the fibulo-talar fibrous tissue. Dorsiflexion improved from 0 degrees to 10 degrees following removal of the exostoses. Post-operatively, active range- of-motion exercises were commenced on the left ankle in order to maintain dorsiflexion and prevent stiffness. The post-operative period was uneventful, and the patient experienced significant resolution in his symptoms. Range of movement in the left ankle joint at follow up at 6 weeks was from 10 degrees of dorsiflexion to 20 degrees of plantar flexion and this motion was maintained until discharge from follow-up. At his one-year follow-up he was pain free and had returned to work and he was discharged from the clinic.

DISCUSSION

A tarsal coalition is a bridging of two or more bones in the hindfoot as a result of failure of mesenchymal segmentation in utero. Only 25% of patients with tarsal coalition are symptomatic making it difficult to accurately assess prevalence of the condition. Calcaneo-navicular and talo-calcaneal are the most common forms of coalition accounting for approximately 90% of cases. In this report, the patient had a talo-calcaneal coalition on the right side in addition to tibio-talar and fibula-talar coalitions on the left. Surgical chilectomy and debridement of the tibio-talar coalition provided symptomatic relief to the patient and enabled his return to work.

As observed in this case talo-calcaeneal coalitions typically occur from the medial aspect of the talus and the sustentaculum tali**.** Key radiological findings are highlighted in this side including talar beak and remodeling of the talo-navicular joint. Both of these indicate chronic loss of supination and pronation in the subtalar joint from the coalition. MRI confirmed loss of joint space at the talo-navicular joint with stress changes and osteophyte formation. Fusion of the degenerate talo-navicular joint on this side resolved the patient's symptoms. We report the first case of failure of mesenchymal segmentation leading to tibio-talar fibrous coalition and fibulo-talar coalition. In this case surgical debridement provided resolution of symptoms.

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Conflicts of Interests: **None**

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