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Bilateral Cubonavicular and Synchronous Talocalcaneal Tarsal Coalition with Stress Response—Case Report and Review of Literature

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Abstract

Tarsal coalition occurs in 1% of the population and represents a congenital failure of segmentation in two or more tarsal bones. It most commonly occurs at the talocalcaneal and calcaneonavicular joint. Although commonly asymptomatic, it may present with pain, rigidity, and pes planus. Cubonavicular, multiple synchronous, and bilateral coalitions are rare but an awareness is required to ensure accurate diagnosis and management. In this article, we presented the first reported case (to the best of our knowledge) of bilateral cubonavicular coalition with synchronous talocalcaneal coalition and stress response within the intermediate cuneiform.

Keywords
- coalition
- tarsal
- synchronous
- stress response

Introduction

Tarsal coalition represents a congenital failure of segmentation in two or more bones of the foot during fetal development. The resultant abnormal union of the involved bones may be osseous (synostosis), cartilaginous (synchondrosis), or fibrous (syndesmosis). It occurs in 1 to 2% of the population and bilaterally in 50% of cases.[1] Talocalcaneal and calcaneonavicular coalitions are the most common forms (collectively 90% of all coalitions), with others, such as talonavicular (third most common), cubonavicular, calcaneocuboid, and naviculocuneiform coalitions being relatively rare.[2,3] Presenting symptoms may include mid or hindfoot pain, stiffness, flatfoot and rigidity, although many patients are asymptomatic. Coalitions are thought to have an autosomal dominant inheritance pattern and may sometimes occur in the context of syndromes, especially when multiple. Outside of syndromal associations, only a few case reports exist describing multiple coalitions in the same foot, with bilateral occurrence being even rarer. Accurate diagnosis of multiple coalitions is essential for treatment decision making. To our knowledge, we present the first reported case of bilateral fibrous cubonavicular coalition associated with unilateral talocalcaneal coalition and intermediate cuneiform stress response, occurring in a 17-year-old male with foot pain.

Case Report

A 17-year-old male patient presented with 18 months' history of insidious onset bilateral midfoot stiffness and pain, worse on the right side. The pain was described as a dull ache that was exacerbated by prolonged standing and

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Fig. 1 Oblique radiograph of the right foot demonstrating irregularity at the articulation between the navicular and cuboid (arrow).

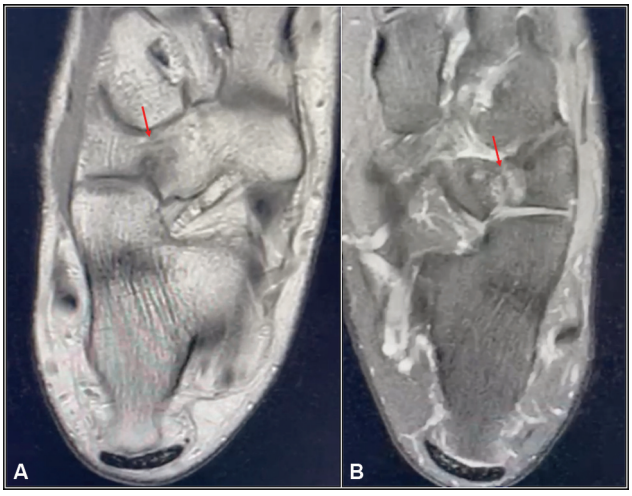

Fig. 2 (**A**) Axial proton density magnetic resonance imaging (MRI) of the left foot and (**B**) axial proton density fat-suppressed-weighted MRI of the right foot demonstrate irregularity and subarticular edema at the articulation between the cuboid and navicular bones, in keeping with fibrous cubonavicular tarsal coalition (arrow).

sporting activities. There was no history of previous trauma, surgical intervention, or other notable medical history. On examination, he demonstrated bilateral pes planus. The overlying skin appeared normal. Dynamic maneuvers of both feet, pronation and supination, were restricted but not painful. There was focal tenderness over the region of intermediate cuneiform of right foot.

Radiographs of both feet were organized to evaluate this further. Weight bearing dorsoplantar view and oblique views were performed. This demonstrated bilateral moderate pes planus with a calcaneal pitch of 5 degrees. There was irregularity of the articulation between the cuboid and navicular bones bilaterally (►**Fig. 1**). In addition, the anterior process of the calcaneum of both feet appeared elongated. The remaining joint spaces were preserved.

Further evaluation was performed with magnetic resonance imaging (MRI). Proton density and proton density fat suppressed sequences in the axial, coronal, and sagittal planes were acquired. MR demonstrated irregularity and mild subarticular edema at articulation between the cuboid and navicular bones bilaterally, in keeping with fibrous coalition (►**Figs. 2** and **3**). A further synchronous fibrous coalition of the subtalar joint, involving the middle facet, was noted on the left ankle (►**Fig. 4**). In the right foot, there was marked osseous edema of the intermediate cuneiform in keeping with a coexisting stress response (►**Fig. 5**). The marrow signal of the rest of the bones was normal. The rest of the joint spaces were preserved with no notable calcaneonavicular coalition.

The patient was managed conservatively. This consisted of an initial period of rest followed by activity modification, analgesia and orthotics.

Discussion

Although tarsal coalitions are often observed as an incidental finding, many patients present with stiffness and pain, which typically subsides with rest, within the mid or hindfoot. Other presentations by ankle sprain, rigidity, flatfoot and peroneal spasticity, secondary to tendon strain from valgus angulations of the foot, have also been reported.[4] Coalitions impede the rotation, motion and gliding normally allowed within a joint, particularly after synostosis. Calcaneonavicular coalition presents earlier than talocalcaneal coalitions, owing to a slightly earlier onset of synostosis, but both present during the adolescent period.[1] The altered foot biomechanics result in an increased load to surrounding structures that may lead to a stress response, stress fracture, or rarely a fracture of coalition.[5,6]

Tarsal coalitions are typically described to have an incidence of 1 to 2%, but retrospective and cadaveric studies have reported higher incidences of 11.5 and 13%, respectively.[7,8] Tarsal coalitions can be difficult to diagnose clinically, owing to nonspecific symptoms. The increased awareness of and contemporary use of MRI may be factors that have increased the detection of coalitions which previously appeared less conspicuous on radiographs or computed tomography (CT). However, even with reformatted cross-sectional imaging, a thorough knowledge of imaging findings, which may be subtle, is required for achieve the correct diagnosis.

The talocalcaneal (subtalar) joint comprises the anterior, middle, and posterior facets. Talocalcaneal coalition most commonly occurs at the middle facet and may occur less commonly in posterior facet, anterior facet, or extra-articular; the latter may be accompanied by an os sustentaculum. They have been classified by Rozansky et al in to five subtypes.[9] Radiographic findings include talar beaking, broadening of the lateral talar process, and narrowing of the posterior talocalcaneal facet. The classic "C sign" can be

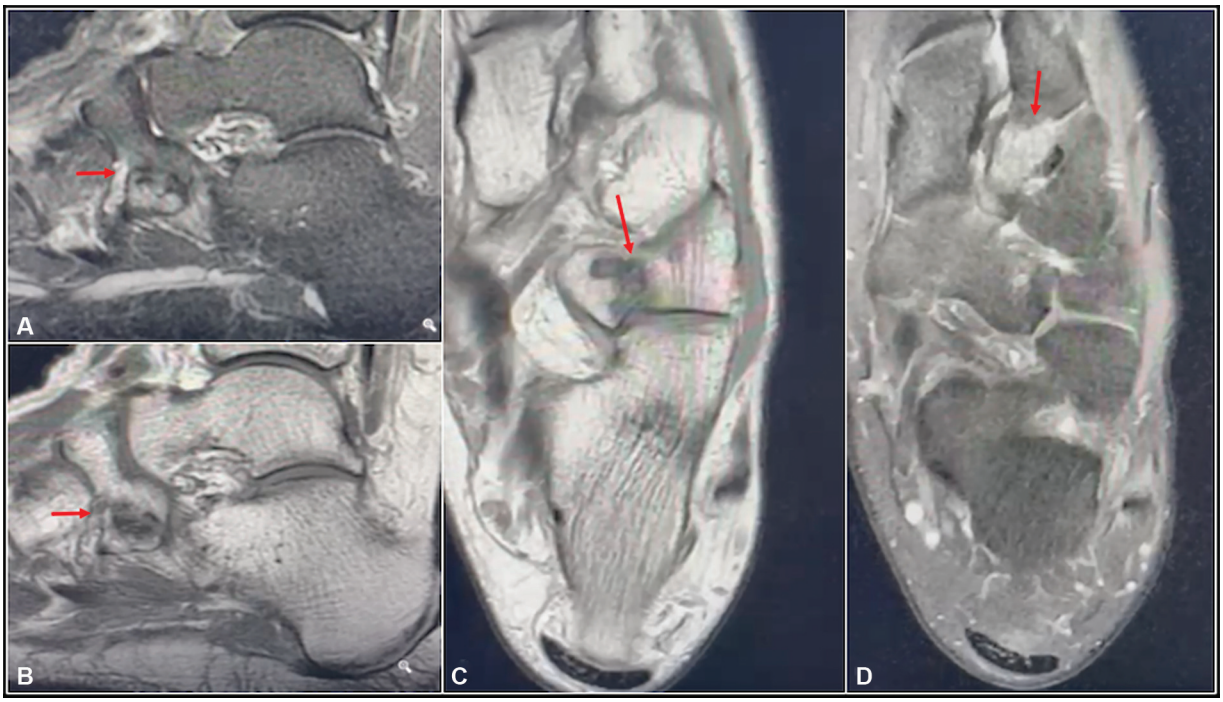

Fig. 3 Imaging of the right foot. (**A**) Sagittal proton density fat-suppressed (PDFS)-weighted, (**B**) sagittal proton density, (**C**) axial proton density, and (**D**) axial PDFS magnetic resonance imaging (MRI) demonstrate irregularity and subarticular edema at the articulation between the cuboid and navicular bones, in keeping with a fibrous cubonavicular tarsal coalition(arrow). (**D**) Axial PDFS MRI also shows marrow edema within the intermediate cuneiform with no discernible fracture line, in keeping with a stress response (arrow).

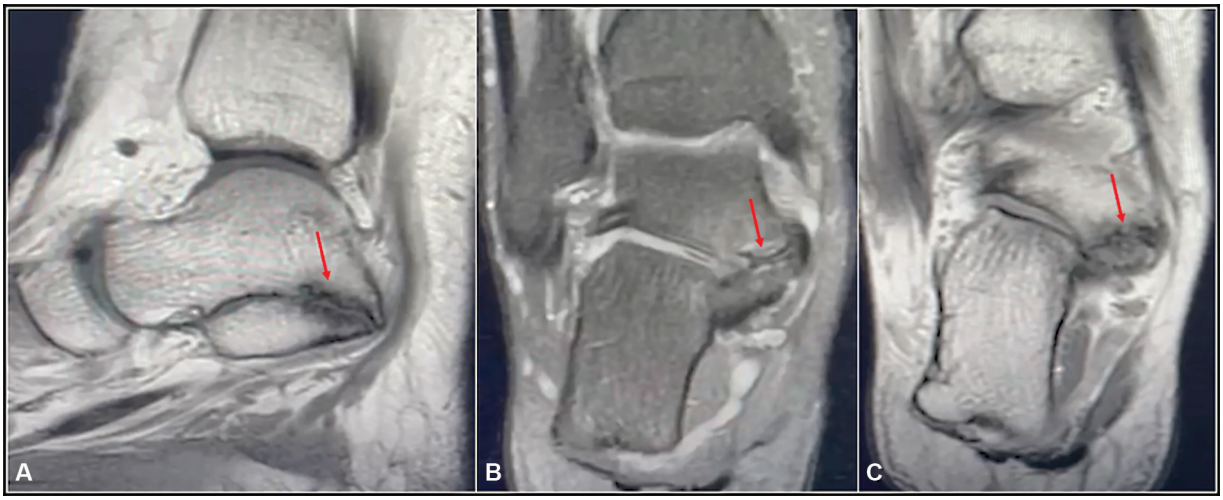

Fig. 4 Imaging of the left foot. (**A**) Sagittal proton density, (**B**) coronal proton density fat suppressed weighted, and (**C**) coronal proton density magnetic resonance imaging demonstrate irregularity, broadening and subarticular edema at the middle subtalar joint facet in keeping with talocalcaneal coalition (arrow).

observed on lateral radiographs but may be absent in non-osseous coalitions or where the sustentaculum tali is hypoplastic.[10] The talocalcaneal coalition may be consequently underreported, when compared with calcaneonavicular coalition, where the anterior calcaneal process ("anteater sign") is more readily appreciated.[3,9] Cubonavicular coalition occurs at the cuboid and navicular articulation. It appears radiographically as an osseous continuity (in synostosis) or with periarticular irregularity (in cartilaginous/fibrous forms). On multiplanar reformatted cross-sectional imaging, tarsal synostosis is identified as a bony continuation of the involved tarsal bones. In cases of synchondrosis and syndesmosis, varying degrees of fibrous/cartilaginous bridging, periarticular irregularity, and subarticular marrow edema are seen.

Talocalcaneal and calcaneonavicular coalitions are quite commonly encountered bilaterally that may owe to a genetic predisposition from autosomal dominant inheritance.[11] Cubonavicular and synchronous (multiple) tarsal coalitions, presented in this case, are less commonly reported with only

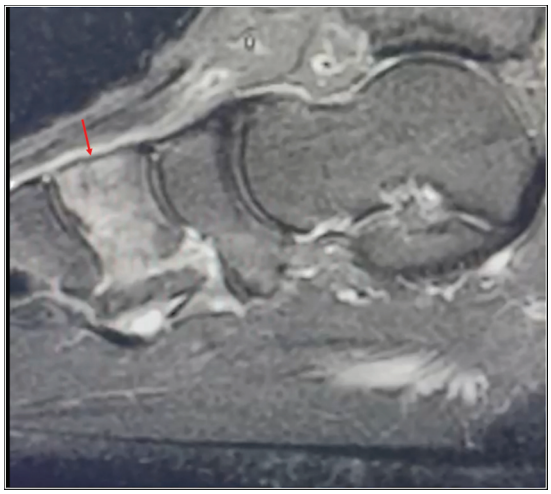

Fig. 5 Sagittal proton density fat suppressed demonstrates marrow edema within the intermediate cuneiform with no discernible fracture line, in keeping with a stress response (arrow).

43 cases in 34 patients reported in the current literature.[12–14] The age at diagnosis ranges from 9 to 47 years old.[15,16] The majority of patients are symptomatic but these data are limited by low case numbers and a probable underreporting in asymptomatic patients. Twelve reported cases demonstrate complete osseous coalition with remainder being cartilaginous or fibrous. Nine cases have been reported to occur bilaterally, with only two of these being nonosseous coalition, as demonstrated in our case.[12,14,17–23]

Multiple coalitions within the same foot are rare. They may not only occur in syndromes such as Apert syndrome, fibular hemimelia, and tarsal–carpal coalition syndrome but also occur outside of syndromic associations.[24,25] Outside of syndromes, there are few case reports of coalitions of the talocalcaneal, calcaneonavicular, and talonavicular joints within the same foot,[25–27] and fewer reporting multiple coalitions occurring bilaterally.[28,29] Farid and Goldcher both reported cases of bilateral "triple coalition" comprising synchronous talocalcaneal, calcaneonavicular, and talonavicular coalition.[30,31] Reddy Mettu et al reported a case of bilateral synchronous tarsal coalition of the talus, calcaneus, navicular, cuboid, cuneiform, and several metatarsals.[32] In 1997, Clark reported six cases of bilateral coalition including one case of bilateral calcaneonavicular with synchronous unilateral cubonavicular coalition.[33] In this case, the cubonavicular coalition was only identified during re-explorative surgery but was present, in retrospect, on the initial CT scan. This case highlights that a reader must avoid satisfaction of search so that, when one coalition is identified, the remaining bones are closely inspected for additional abnormalities.[34,35]

In our patient, we also observed a stress response coexisting with cubonavicular coalition in the left foot. It is thought that ossification of an existing coalition may be the cause of pain experienced by patients but recent cases have revealed multiple cases with coexisting tarsal stress injuries. Stress injuries occur from overuse secondary to repetitive loading that can be exaggerated by altered foot biomechanics. In 2021, Jain et al presented six cases of tarsal coalition with coexisting stress injuries or fractures and proposed that these entities may be related.[6] Stress response and fracture have been described by several other authors in cases of tarsal coalition,[36,37] with our case being the first to make this observation in a patient with cubonavicular coalition. MRI has an excellent sensitivity in identifying stress injuries with the additional benefit of better differentiating between subtypes of tarsal coalition.[38]

In symptomatic tarsal coalition, the first-line therapy would typically include conservative therapy with orthotics, activity modification, analgesia, and rest. In cases where nonoperative treatment has failed, surgery by means of resection, interposition, or fusion of the affected tarsal bones may be required. Close clinical and radiological follow-up is required to avoid secondary stress injuries, fractures, and arthritis.[12]

Although it is well known that coalitions frequently involve both feet, multiple synchronous and bilateral tarsal coalition are a rare and less well-recognized entity. It may be a source of chronic and sometimes relentless foot pain in adolescent patients. When assessing coalitions, physicians and radiologists must be aware of synchronous coalitions (those other than calcaneonavicular and talocalcaneal), stress responses, familial predilection, and possible syndrome association. Accurate diagnosis is essential to guide correct decision making for management.

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Conflict of Interest
None declared.

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