

Cubonavicular Coalition in a Middle-Aged Woman

A Case Report

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Cubonavicular coalition is a rare type of tarsal coalition that can be described as osseous or nonosseous (fibrous, cartilaginous, or fibrocartilaginous). Typically, it manifests symptoms during adolescence, as it presents with pain at the Mid-hindfoot and with decreased range of motion at the midtarsal joints, hindfoot valgus deformity, or peroneal spasm. Here, we present a rare case of cubonavicular coalition in a middle-aged woman with atypical presentation and a review of the literature. We conclude that this abnormality should be taken into account in the differential diagnosis of mid-hindfoot pain, even in middle-aged adults. (J Am Podiatr Med Assoc 112(6), 2022)

A rare cause of pain in the midfoot is tarsal coalition. Tarsal coalitions are rare abnormalities, accounting for less than 1% of all tarsal conditions, when two or more tarsal bones are united and may be osseous (synostosis) or nonosseous (fibrous syndesmosis, cartilaginous synchondrosis, or fibrocartilaginous).¹ However, the actual incidence of the condition is in fact underestimated because of a subset of patients in the general population being asymptomatic.² The most common types of tarsal coalitions are talocalcaneal and calcaneonavicular, representing more than 90%.³ Furthermore, tarsal coalitions are intra-articular (bridge) or extra-articular (a bar), with cubonavicular coalition being an extra-articular type and talocalcaneal being an intra-articular type.⁴

Cubonavicular coalition is an uncommon type of tarsal coalition and a rare cause of pain in the

midfoot, which is usually encountered among adolescents or young adults.⁵ Most of the time, it is congenital, usually transmitted in an autosomal dominant or autosomal recessive fashion, because of a failure of differentiation of the primitive mesenchyme in the first periods of intrauterine development, and it can be correlated with other abnormalities such as symphalangism and clinodactyly.^{6,7} Occasionally, cubonavicular coalitions can be acquired after trauma, infection, arthritis, and tumor.⁸ More than 50% of the patients have bilateral cubonavicular coalition.⁹

Typical clinical presentation includes chronic aching pain of the midfoot or hindfoot exacerbated during ambulation, with or without a history of ankle injury.^{10,11} However, it could also stay asymptomatic, as Williamson and Torode¹² initially reported in a bilateral case of cubonavicular coalition. Clinical examination may reveal decreased range of motion at the midtarsal and subtalar joints, tenderness and swelling over the site of coalition, hindfoot valgus deformity, and peroneal spasm.¹³⁻¹⁵ The analysis of cubonavicular coalition cases in the literature revealed that symptoms were more frequent for non-osseous than in osseous unions, but the limited number of these cases does not allow us to draw any certain conclusions.¹⁶ In the adult population, asymptomatic coalitions usually become symptomatic after a minor trauma.¹⁷

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Case Report

A 50-year-old woman with a medical history that included hypertension and hyperlipidemia, and without a surgical history, presented to our orthopedic department complaining of chronic pain in her left foot at the midtarsal joint area. The patient's symptoms started 4 months before the visit with an insidious onset and have been escalating ever since, especially while ambulating, with the pain during past week being described as intense, burning, and constant that occasionally caused sleep disruption. Pain was assessed using the visual analog scale (VAS) and was rated by the patient as 9. She had no history of previous trauma or rheumatologic disease, or family history of similar complaints. Physical examination of the left lower extremity showed tenderness on palpation of the dorsum of the foot, normal range of motion, but painful dorsiflexion of the foot. No signs of deformity or appreciable swelling were observed. Plain anteroposterior and lateral oblique radiographs of the left foot were ordered, which were negative for soft-tissue injury, fracture, or dislocation, and showed no other specific pathologic lesion (Fig. 1). Initially, the patient was treated conservatively, nonsteroidal anti-inflammatory drugs were prescribed, the ankle joint was immobilized in a short leg posterior splint, and partial weightbearing was allowed. After 1 week, no signs of improvement were observed and a magnetic resonance imaging (MRI) scan of the left foot for further radiologic assessment was obtained, which revealed the presence of cubonavicular coalition (Fig. 2). Cortisone plus lidocaine injection into the site of coalition under ultrasound guidance was performed (Fig. 3)

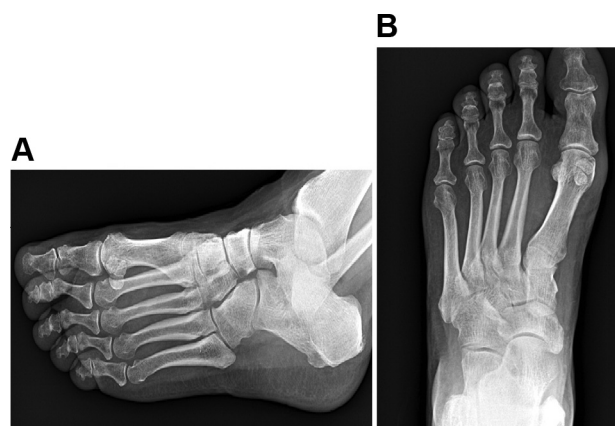


Figure 1. Lateral oblique (A) and anteroposterior (B) radiographs without specific findings of cubonavicular coalition.

and led to immediate resolution of symptoms, indicating that the cubonavicular coalition was the sole cause of pain. Full weightbearing as tolerated in a walking boot was allowed for an additional week. At reevaluation, the patient was asymptomatic, had a VAS score of 0, and could ambulate pain free, so the walking boot was removed. At 6- and 9-month follow-up, the VAS score remained 0, there was no recurrence of symptoms, and no further therapeutic measures were required.

Discussion

Coalition between cuboid and navicular bones is extremely rare, accounting for less than 1% of all tarsal coalitions, and usually becomes symptomatic during adolescence, severely affecting the quality of life and function of all patients.^{14, 18, 19} However, in our case, this condition presented in a middle-aged woman, which is extremely rare, as there are only nine cases from the available data reported in the literature.¹⁸ Moreover, our case had an atypical clinical presentation, without hindfoot valgus deformity and peroneal spasm.

Radiographic findings depend on the type of the coalition, and usually plain radiographs are sufficient for the diagnosis of the disorder.²⁰ In the case of osseous coalition, anteroposterior and oblique views will reveal a bony bar between cuboid and navicular bones, whereas in the nonosseous type, joint space narrowing and bony margin irregularities are the main findings.²¹ Advanced imaging can be performed in cases of inconclusive radiographs or when surgical planning is needed. Computed tomographic imaging will demonstrate a solid bone bridging between the two bones in the osseous form of coalition or abnormal orientation and narrowing of the joint space and reactive bone changes in the case of the nonosseous form.¹⁷ On MRI of cubonavicular coalitions, bone marrow edema adjacent to the abnormal joint is often identified.¹⁷ Osseous coalitions demonstrate continuation of bone marrow signal across the fused bones; cartilaginous coalitions demonstrate intermediate T1 signal and intermediate to hyperintense T2 signal across the joint; and fibrous coalitions demonstrate low signal intensity on all pulse sequences.¹⁶ In our case, although radiographic findings were negative for cubonavicular coalition, MRI was typical for a nonosseous type.

The treatment of cubonavicular coalition can be either conservative or operative. Nonoperative treatment is the first-line therapy and the reasonable

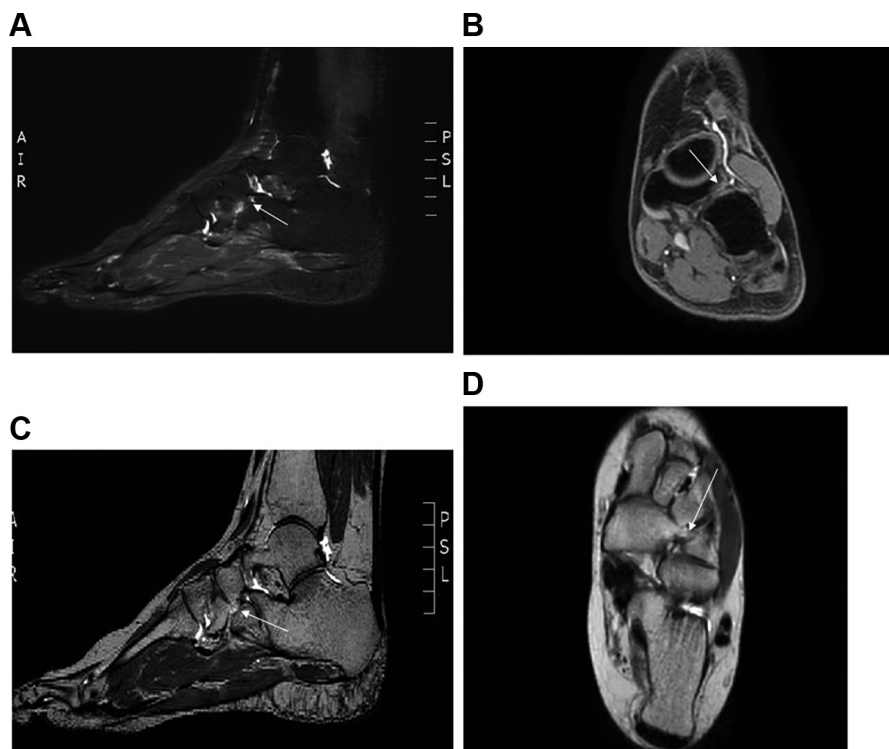


Figure 2. (A and B) Sagittal and axial T1-weighted magnetic resonance images demonstrating bone marrow edema adjacent to the joint and bony margin irregularities between the cuboid and navicular bones (white arrows). (C and D) Sagittal and axial T2-weighted magnetic resonance images demonstrating a fibrocartilaginous bar of intermediate hyperintense signal between the cuboid and navicular bones (white arrows).

option at initial presentation.^{13,18,19} Conservative treatment consists of rest, cold compresses, nonsteroidal anti-inflammatory drugs, short-leg splint, ankle

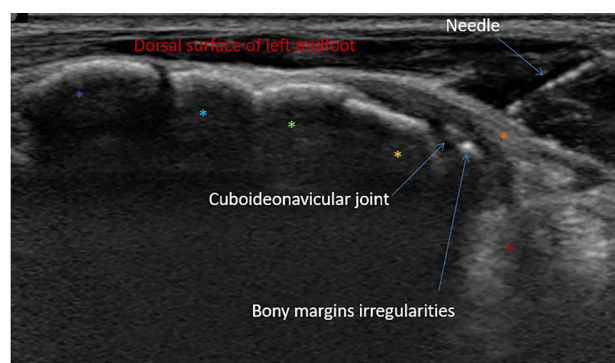


Figure 3. Short axis view ultrasound image of the left midfoot. A corticoid injection is performed at the site of the cubonavicular coalition. Bony margin irregularities of cuboideonavicular joint are evident. Dorsal Cubonavicular ligament (orange asterisk), Navicular (yellow asterisk), Cuboid (red asterisk), Medial Cuneiform (purple asterisk), Intermediate cuneiform (blue asterisk), Lateral cuneiform (green asterisk).

braces, orthotics (heel lifts), physiotherapy, and steroid injections with or without local anesthetic.^{4,13,15} Surgical treatment is considered for patients who have deformities such as peroneal spasm or valgus hindfoot, for osteoarthritic changes, for patients in whom the conservative treatment has failed and have consistent pain, and for unresected or malresected coalitions.^{8,13,20} The surgical intervention includes resection of the coalition with or without the interposition of autogenic or allogenic grafts (eg, adipose tissue, tendon, tensor fascia lata, superficial fascia, bone wax, cryopreserved amniotic membrane with autogenous adipocyte graft).⁴ The preferred approach for the resection is the dorsolateral incision over the superior aspect of the cuboid.¹⁸

For severe cases of cubonavicular coalition in which there are severe degenerative changes, multiple joint coalitions, or when the previous resections have failed, subtalar or triple arthrodesis of the involved joints is recommended.^{4,13,18} In adolescents and young adults, after failure of the conservative measures, surgical intervention with resection of the coalition to restore hindfoot motion and alleviate symptoms is recommended even without radiographic evidence

of degenerative joint disease or associated tarsal coalitions in other joints.¹⁸ Taking into account that in our case the criteria for operative treatment were not met, we elected for conservative treatment, which was proven to be successful until the patient's last visit to our orthopedic department.

Conclusions

In summary, cubonavicular coalition is a rare cause of midfoot or hindfoot pain that should be considered in the differential diagnosis not only in adolescents or young adults but also in middle-aged patients, as in our case. Although cubonavicular coalition was initially described in 1957 by Waugh¹⁰ and in 1959 by Del Sel and Grand,²¹ it has since been poorly reported and warrants further characterization to provide the best evidence-based management. Importantly, cubonavicular coalition can present with atypical clinical findings and no history of trauma, similar to our case. Plain radiographs can be inconclusive; in that case, advanced imaging with computed tomographic scans or MRI should be used.¹¹ Conservative treatment is the first-line therapy, with better results in adults than in adolescents, who very often require surgical management.²²

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