

Cryopreserved Amniotic Membrane and Autogenous Adipose Tissue as an Interpositional Spacer Following Resection of a Cubonavicular Coalition: A Case Report and Literature Review Duane J. Ehredt, Jr, DPM, FACFAS¹; Eli G. Elias, DPM²; Emily Zulauf, BS³; Svetlana Motorikina, BS³; James Connors, DPM, AACFAS¹

Statement of Purpose

The purpose of this poster is to present a novel, and previously undocumented use for cryopreserved amniotic membrane with autogenous adipose tissue interposition for the treatment of a cubonavicular coalition in an adult male. Cubonavicular coalitions are exceedingly rare, and a literature review is presented to the reader as well.

Literature Review

Tarsal coalition, a rare abnormality of union between two or more tarsal bones, affects approximately 1-2% of the general population [1,2, 3]. It may be classified as congenital or acquired, congenital being the most common [4]. It may also be classified according to the type of union: osseous synostosis, fibrous syndesmosis, cartilaginous synchondrosis or a combination of these types. Additionally, coalitions may be categorized as intra-articular, a bridge, or extra-articular, a bar. Roughly 90% of reported tarsal coalitions are located between the talus and calcaneus (talocalcaneal bridge) or calcaneus and navicular (calcaneonavicular bar). Less than 1% of tarsal coalitions are reported as occuring between the cuboid and navicular, also referred to as a cubonavicular or cuboid-navicular coalition. By definition, a cubonavicular coalition is an extra-articular bar, and Waugh first described it in 1957 in association with peroneal spastic flatfoot [5]. In 1991, Palladino et al reported only 17 cases of cubonavicular coalition ever being described in medical literature worldwide [6].

Classically, tarsal coalitions become symptomatic during adolescence when the coalition undergoes ossification and the bones are subject to repeated mechanical stress and remodeling. Patients will typically present with unilateral or bilateral foot pain, described as deep and aching in nature [4], which is exacerbated with activity, increase in weight, or walking on uneven surfaces [7]. Bilateral tarsal coalitions have been described in over 50% of cases [8].

Clinically, a tarsal coalition may present as limitation or loss of motion at the subtalar joint. Cubonavicular coalitions; however, may present with limited mobility to midtarsal joint inversion, with preservation of motion at the subtalar joint [3]. Symptoms of a cubonavicular coalition can be similar to, and thus must be distinguished from, calcaneonavicular coalitions. Asymptomatic presentations of tarsal coalition have been reported in the literature; however, clinical presentation of pain, limitation of motion, and muscle spasm should increase suspicion for tarsal coalition [4].

Radiographic evaluation of standard plain films imaging can be sufficient to accurately diagnose a tarsal coalition. Plain film radiographs have been found to be 88% specific for talocalcaneal and 97-98% specific for calcaneonavicular coalitions [9]. Cubonavicular coalitions are best viewed on lateral or medial-oblique radiographs. Advanced imaging is typically reserved for inconclusive radiographs or surgical planning for coalition resection [4]. MRI is frequently utilized to evaluate cubonavicular coalitions.

Evidence based medicine for diagnosing and treating cubonavicular coalitions have yet to be defined in modern medical literature. In 1978, Cavallaro and Hadden described the first detailed surgical approach for resection of a cubonavicular bar [10]. One and two incision approaches have also been described [4]. Multiple authors have suggested various interpositional grafts as adjuncts to surgical resection. In this case report, we present the novel use of cryopreserved amniotic membrane with autogenous adipocyte interposition, along with resection of a cubonavicular coalition. To our knowledge, this is the first known attempt at this treatment strategy with greater than 12 months of follow up.

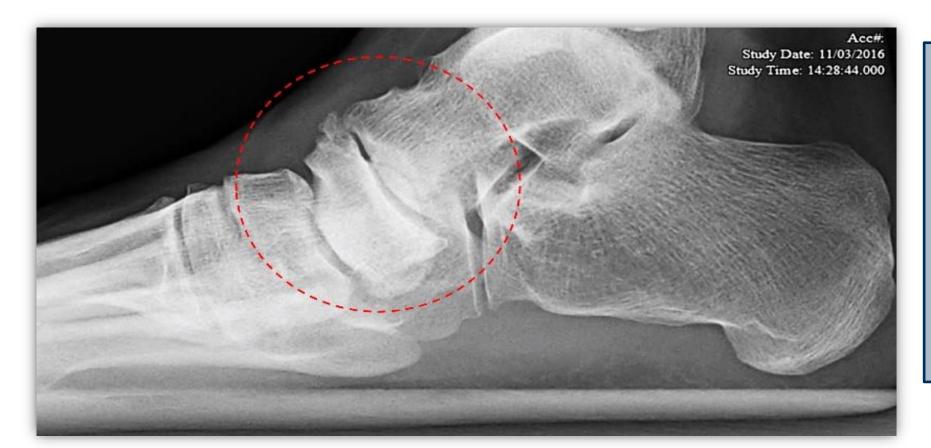


Figure 1. Plain film radiography with both primary and secondary signs of cubonavicular coalition present (dotted red circle)

Case Study

A 34-year old male presented to the senior author's (DJE) clinic with a history of chronic aching pain localized to his left dorsolateral midfoot. His pain began insidiously approximately two years prior and he denied any particular traumatic event. He did acknowledge a heavy ambulatory demand for his job requirements as a warehouse laborer. He described the initial pain as dull and continuously aching which later progressed to sharp and excruciating pain during ambulating and with extended periods of activity. His past medical and surgical history were unremarkable. Initial conservative treatment, including nonsteroidal anti-inflammatory oral medications, a pneumatic offloading boot, orthotic inserts, and corticosteroid injections, provided only minimal short-term pain relief.



Figure 1 continued.

A focused physical examination of the left lower extremity demonstrated a hindfoot valgus deformity, complete loss of the medial longitudinal arch, and decreased range of motion at the midtarsal and subtalar joints, indicative of a rigid pes plano-valgus deformity. Three weightbearing plain-film radiographic views (dorsoplantar, medial oblique and lateral) were obtained of the left foot, and were negative for soft tissue injury, fracture and dislocation. Imaging was suggestive, however, for a cubonavicular coalition [Figure 1]. Magnetic resonance imaging was performed and revealed a fibro osseous coalition between the cuboid and the lateral aspect of the navicular [Figure

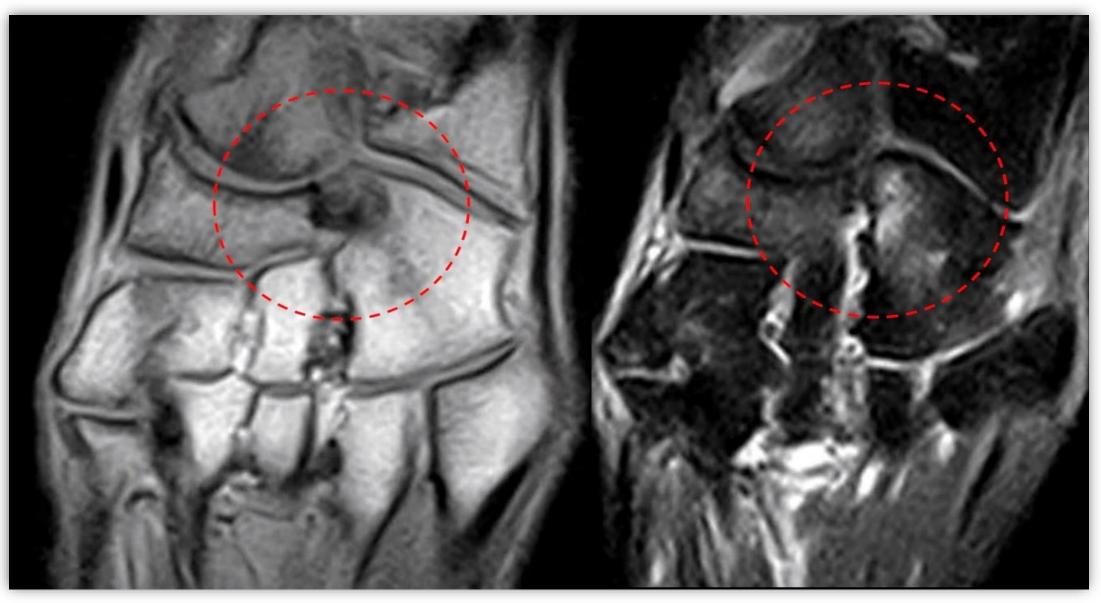


Figure 2. Both T1 and T2 weighted MRI demonstrating fibro-osseous cubonavicular coalition

The progressive worsening of the patient's symptoms in conjunction with failure of conservative treatments led to the discussion of operative intervention. Hindfoot arthrodesis and coalition resection with interposition were discussed in detail with the patient. Given his age, deformity, and secondary signs of arthrosis the senior author favored hindfoot arthrodesis; however, the patient voiced concern regarding losing his job due to the prolonged non-weight bearing period post-operatively. Reported surgical outcomes of coalition resection in older patient populations were discussed with the patient, specifically the increased risk for recurrent joint limitation and return of previous symptoms [11]. Ultimately, the patient elected to proceed with surgical resection of the coalition with interpositional graft placement, with the understanding that eventual hindfoot arthrodesis may be necessary.

Surgical Technique

The patient was positioned supine on the operating table with a sandbag placed under the ipsilateral hip to internally rotate the operative extremity. Hemostasis was achieved and maintained throughout the case with a pneumatic thigh tourniquet set to 300 mmHg. Under fluoroscopic guidance, osseous anatomical landmarks were mapped with a skin marker. A curvilinear incision was made from the calcaneocuboid joint/ sinus tarsi region to the distal aspect of the articulation between the cuboid and navicular bones [Figure 3]. The dissection was deepened in anatomical fashion through the incision site. A peroneus tertius tendon was identified and retracted superiorly for the remainder of the case. The extensor digitorum brevis (EDB) muscle belly was identified, bluntly dissected free from the overlying deep fascia, and transected from its origin at the anterolateral floor of the sinus tarsi and anterior process of the calcaneus. Next, the EDB muscle belly was dissected from the lateral talocalcaneal joint and from the articulation between the cuboid and navicular. At this time, the fibro-osseous coalition between the cuboid and navicular bones was visualized and confirmed with fluoroscopic assistance.



Figure 3. Curvilinear incision for access to the cubonavicular coalition. Note the secondary dorsal incision which was ultimately not needed

Sharp osteotomes were then utilized to completely resect the coalition [Figure 4]. A 6mm wedge of tissue consisting of bone from the navicular, fibro-osseous coalition, and bone from the cuboid was extirpated from the left foot. Following resection, an increase in motion of the midfoot complex was appreciated in all three cardinal planes. Complete coalition resection was confirmed using fluoroscopy [Figure 5]. A reciprocating rasp was then utilized to smooth remaining sharp osseous edges and a needlepoint Bovie was used to cauterize the bleeding bone.

Figure 4. The cubonavicular coalition was identified clinically and radiographically. The surgeon uses an osteotome to perform the 6mm resection

Approximately 3cc of subcutaneous fat harvested from the sinus tarsi ("Hoke's Tonsil") and was wrapped in a 3cm x 3cm cryopreserved amniotic membrane allograft prior to being interposed between the cuboid and navicular in the area of osseous resection. The combination of adipocyte autograft with amniotic membrane allograft is novel and previously undocumented in the literature. The amniotic membrane allograft was utilized to supplement the interpositional mass in effort to decrease recurrence [12, 13] as well as to reduce scarring and modulate inflammation. Following a saline flush, the EDB muscle belly was re-approximated and the wound was closed in anatomical layers. The closed incision was then dressed with bacitracin ointment, a non-stick sterile dressing, and a two-layer Jones compression bandage with a posterior splint.

The post-operative course consisted of two weeks of non-weight bearing in the posterior splint followed by two weeks of full weight bearing as tolerated in a CAM walking boot. The patient completed a formal eightweek course of physical therapy with emphasis on hindfoot strength and range of motion exercises. At eight weeks post-operatively, the patient was permitted to return to work without restrictions.





At post-operative follow-up of 12 months, the patient demonstrated maintenance of an increased range of motion of the midfoot in the transverse, sagittal and frontal planes and was ambulating without pain. He is currently ambulating in normal shoe gear with over-thecounter inserts for arch support.

> Figure 5. Final radiographic images confirm cubonavicular coalition resection. Improvement in midfoot and hindfoot range of motion was appreciated.



Analysis & Discussion

Tarsal coalitions result from abnormalities in differentiation in the first stages of development [14]. A genetic correlation has been described as well as combined presentation of tarsal coalition with hereditary symphalangism and clinodactyly [15]. Classic presentation of tarsal coalition is more common in males than in females and is often described by patients as progressive pain and stiffness in the midfoot and hindfoot [16]. Clinically, midfoot and hindfoot range of motion is decreased. Radiographs may be sufficient for diagnosis of tarsal coalitions; however, in the case of cubonavicular coalition, advanced imaging may be warranted. To diagnose cubonavicular coalition with radiographs, one must anticipate subtle changes on imaging in the area between the cuboid and navicular. Abnormal weight-bearing mechanics associated with other tarsal coalitions are not visible on radiographs in the case of a cubonavicular coalition [16] making diagnosis with radiography more challenging in some cases. It is the authors' opinion that magnetic resonance imaging in conjunction with plain film radiography is necessary for accurate diagnosis of cubonavicular coalition.

Conservative treatment of symptomatic coalitions may consist of non-steroidal anti-inflammatory medications, short leg casting, steroid injections and orthotics [16]. Historically, tarsal coalitions failing conventional therapy have been treated with resection with interposition of some medium: autogenic/allogenic grafts (adipose tissue, tendon, tensor fascia lata, superficial fascia), bone wax or nothing [12,13]. For patients who fail conservative treatment, surgical intervention in the form of coalition resection with graft interposition may be indicated, and has been demonstrated to be an effective treatment option [17].

Cubonavicular coalition is very rare and is, therefore, not well documented in the literature. In any patient presentation of chronic midfoot pain, differential diagnoses should include tarsal coalitions, notably cubonavicular coalition. A high index of suspicion should be present for cubonavicular coalition on radiographic examination of plain films. While conservative treatment may be an effective remedy for symptomatic coalition, surgical intervention should be considered in cases of chronic midfoot pain failing conventional therapies.

In conclusion, cubonavicular coalitions are a rare congenital anomaly that can cause debilitating midfoot and hindfoot pain. Currently there is no well documented consensus for treatment. This case report details our approach to cubonavicular coalition resection, and interposition with cryopreserved amniotic membrane and autogenous adipocyte graft. The goal of this procedure was to reduce pain and inflammation, while improving foot function. Although longer follow up and multiple cases will be necessary for a more formal evaluation of this therapy, we believe this is a viable surgical option for treatment of the symptomatic cubonavicular coalition.

References

- . Newman J.S., Newberg A.H. "Congenital tarsal coalition: multimodality evaluation with emphasis on CT and MR imaging," Radiographics. 2000;20:321–332.
- 2. Crim J. Imaging of tarsal coalition. Radiologic clinics of North America. 2008 Nov 30;46(6):1017-1026. 3. Prado MP, Mendes AA, Olivi R, Amodio DT. Cuboid-navicular tarsal coalition. Revista Brasileira de Ortopedia (English Edition). 2010 Oct 31;45(5):497-499.
- 4. McGlamry's Comprehensive Textbook of Foot and Ankle Surgery, Fourth Edition. Lippincott Williams & Wilkins. 2012;598-632.
- 5. Waugh W. Partial cubo-navicular coalition as a cause of peroneal spastic flat foot. Bone & Joint Journal. 1957 Aug 1;39(3):520-523
- 6. Palladino SJ, Schiller L, Johnson JD. Cubonavicular coalition. Journal of the American Podiatric Medical Association. 1991 May;81(5):262-266.
- 7. Wright EM, Green D. Tarsal coalition. Banks AS, Downey MS, Martin DE, Miller SJ. McGlamry's Comprehensive Textbook of Foot and Ankle Surgery. 1997;1:999-1032. 8. Ehrlich M.G., Elmer E.B. Tarsal coalition. In Disorders of the Foot and Ankle, 2nd edition. Jahss, M. (ed.), Philadelphia, W.B. Saunders, 1991, 921-938.
- 9. Crim JR, Kjeldsberg KM. Radiographic diagnosis of tarsal coalition. American Journal of Roentgenology. 2004 Feb;182(2):323-8.
- 10. Cavallaro DC, Hadden HR. An unusual case of tarsal coalition: a cuboid navicular synostosis. Journal of the American Podiatric Medical Association. 1978 Feb;68(2):71-5.
- 11. Downey M.S. Keys to Treating Tarsal Coalitions. Podiatry Today 2011;24:48-56. 1
- 12. De Wouters S, Duy KT, Docquier PL. Patient-specific instruments for surgical resection of painful tarsal coalition in adolescents. Orthopaedics & Traumatology: Surgery & Research. 2014 Jun 30:100(4):423-7.
- 13. Raikin S, Cooperman DR, Thompson GH. Interposition of the split flexor hallucis longus tendon after resection of a coalition of the middle facet of the talocalcaneal joint. JBJS. 1999 Jan 1;81(1):11-9.
- 14. Mubarak SJ, Patel PN, Upasani VV, Moor MA, Wenger DR. Calcaneonavicular coalition: treatment by excision and fat graft. Journal of Pediatric Orthopaedics. 2009 Jul 1;29(5):418-26. 15. Mata SG, Hidalgo-Ovejero A. Cuboid-navicular tarsal coalition in an athlete. InAnales del sistema sanitario de Navarra 2011 May 1 (Vol. 34, No. 2, pp. 289-292). 16. Awan O, Graham JA. The rare cuboid-navicular coalition presenting as chronic foot pain. Case reports in radiology. 2015 Jan 26;2015.
- 17. Sarage AL, Gambardella GV, Fullem B, Saxena A, Caminear DS. Cuboid-navicular tarsal coalition: report of a small case series with description of a surgical approach for resection. The Journal of Foot and Ankle Surgery. 2012 Dec 31;51(6):783-6.