Case Reports

Symptomatic Bilateral Talonavicular Coalition

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Abstract Congenital talonavicular coalition is reported less frequently than talocalcaneal or calcaneonavicular coalition and represent approximately 1% of all tarsal coalitions. Although reportedly transmitted as an autosomal-dominant disorder, tarsal coalition may be inherited as an autosomal-recessive trait. It has been associated with various orthopaedic anomalies, including symphalangism, clinodactyly, a great toe shorter than the second toe, clubfoot, calcaneonavicular coalition, talocalcaneal coalition, and a ball-and-socket ankle. Patients with talonavicular coalitions are usually asymptomatic and rarely undergo surgical treatment. We report the case of a 24-year-old woman with symptomatic bilateral talonavicular coalitions and previously unreported associated anomalies (nail hypoplasia and metatarsus primus elevatus) and review the relevant literature. The patient underwent surgery (calcaneocuboid joint distraction arthrodesis and a proximal plantar flexion osteotomy with a dorsal open wedge of the first metatarsal). At 1-year followup, she was pain-free with better alignment of both feet and showed

radiographic consolidation of the arthrodesis. Although this condition is less likely to be clinically important than other tarsal fusions, it sometimes can be painful enough for the patient to undergo surgery.

Introduction

The congenital anomaly talonavicular coalition is reported less frequently than talocalcaneal or calcaneonavicular coalition [2, 4, 11]. The relative incidence of tarsal coalitions is reportedly 1.3% of all tarsal coalitions. Although reportedly transmitted as an autosomal-dominant disorder [1, 2, 4, 7, 11], tarsal coalition may be inherited as an autosomal-recessive trait [11]. It has been associated with various orthopaedic anomalies, including symphalangism [1–4, 7], clinodactyly [2, 4], a great toe shorter than the second toe [4, 11], clubfoot [4], and a ball-and-socket ankle [4, 6]. Patients with talonavicular coalitions are usually asymptomatic [5, 7, 11] and rarely undergo surgery.

We report the case a 24-year-old woman with bilateral symptomatic talonavicular coalition associated with bilateral nail hypoplasia of the fourth and fifth toes, anomalies that have not been described previously in individuals with talonavicular coalition, and review the relevant literature.

Case Report

A 24-year-old woman reported a 19-year history of pain and fatigue after prolonged standing or sports activities. Her symptoms were not continuous. The patient had been treated nonoperatively occasionally with orthotics and physiotherapy since the age of 5 years without improvement. Physical examination revealed full ankle and

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subtalar motion. The patient had mild midfoot hyperpronation (Fig. 1A) and painful protuberances over the medial border of each foot (Fig. 1B) and also in the dorsal aspect of the first metatarsal head bilaterally (metatarsophalangeal joint). Both great toes had full range of motion.

Fig. 1A–B (A) A posterior-view preoperative photograph shows mild hyperpronation, and (B) the anterior view shows the medial bone lump.

Fig. 2 The patient's pedigree is shown. The previously unreported symptoms include nail hypoplasia and metatarsus primus elevatus.

She had nail hypoplasia in the fourth and fifth toes of both feet. The patient's family also was evaluated to assess for a genetic pattern of transmission (Fig. 2). Her father had an asymptomatic bilateral talonavicular coalition and bilateral fifth finger clinodactyly in his hands. Her younger sister also had an asymptomatic bilateral talonavicular coalition.

Weightbearing radiographs showed bilateral talonavicular fusion (Fig. 3A–C). On the lateral view, a large space at the navicular-cuneiform joint could be seen (Fig. 3B). It also revealed a metatarsus primus elevatus. On the anteroposterior view, we observed an increased talo-first metatarsal angle. MRI did not show any additional anomalies.

To confirm the joint as the source of her pain, we injected her calcaneocuboid with lidocaine and the patient had almost complete pain relief for 2 hours, but her symptoms then returned.

We operated on the right foot first and then on the left foot 1 year later. The procedures were a calcaneocuboid joint distraction arthrodesis with a tricortical autograft (obtained from the iliac crest) fixed with an H plate with 3.5-mm screws, a proximal plantar flexion first metatarsal osteotomy with a dorsal open wedge (autograft) fixed with a 2.7-mm screw, and a percutaneous Achilles tendon lengthening. Each time the patient's foot was immobilized in a cast for 1 month and kept nonweightbearing for 2 months. Physiotherapy was performed until full weightbearing at 3.5 months.

One year after the second surgery, she had achieved pain relief and had good alignment of her feet. Subtalar motion was full. Radiographs showed consolidation of the arthrodesis (Fig. 4). The medial bone lump had disappeared and the hyperpronation had improved (Fig. 5). Clinical improvement of elevation of the first metatarsal was not observed on the postoperative radiographs.

Fig. 3A–C (A) A preoperative weightbearing radiograph (dorsoplantar view) shows the talonavicular synostosis in the right foot. (B) A lateral view shows a large space at the navicular-cuneiform joint and metatarsus primus elevatus. (C) An oblique view shows the talonavicular synostosis.

Fig. 4A–B Postoperative (A) lateral and (B) anteroposterior radiographs show consolidation of the arthrodesis at the calcaneocuboid joint and of the first metatarsal osteotomy.

Fig. 5A–B (A) An anterior-view postoperative photograph shows disappearance of the medial bone lump, and the (B) posterior view shows improvement of the hyperpronation.

Table 1. Relative incidence of talonavicular coalition

Study	Number of cases	Symptoms	Associated anomalies	Treatment
Bonk and Tozzi [1]	1	Yes	No	Nonoperative
Challis [2]	4	Yes (1); no (3)	No	Nonoperative (3); operative (1)
David et al. [3]	1	Yes	No	Nonoperative
Doyle and Kumar [4]	3	Yes	Yes	Nonoperative
Frost and Fagan [5]	1	Yes	No	Nonoperative
Kramhøft and Monberg [6]	1	No	No	Operative
Lahey et al. [7]	6	Yes (4); no (2)	No	Nonoperative (4); operative (2)
Pontious et al. [9]	1	Yes	No	Nonoperative
Stormont and Peterson [10]	4	No	Yes	Nonoperative (3); operative (1)
Zeide et al. [11]	4	Yes (1); no (3)	Yes	No
Current authors	1	Yes	Yes	Operative

Discussion

In 1879, Anderson recorded the first case of talonavicular coalition in an anatomic dissection [4]. Small series [7, 11] and isolated case reports [1–3, 6, 9] have been published and several have reported a genetic transmission [1, 7] and an association with various orthopaedic anomalies [1–3, 6, 7, 11] (Table 1).

Several theories have been offered regarding the etiology underlying this rare anomaly. Failure of differentiation and segmentation of the primitive mesenchyme was described as the etiology of talonavicular coalitions [1, 3, 6, 9]. In our patient, a positive family history reveals the coalition to be familial. The presence of clinodactyly and talonavicular fusions in some of the patients' relatives suggests a variable expression and dominant inheritance that may occur in some families.

Previous publications [2, 6] have reported this coalition is associated with several orthopaedic anomalies, including symphalangism [1–4, 7], clinodactyly [2, 4], a great toe shorter than the second toe [4, 10], clubfoot [4], and a ball-and-socket ankle [4, 6]. In our patient with bilateral anomalies, we observed a metatarsus primus elevatus and nail hypoplasia; we found no reports of these associations.

Most calcaneonavicular and talocalcaneal coalitions present as a painful foot of some type [1–4, 6, 8, 10]. In contrast, patients with talonavicular synostosis usually are asymptomatic [5, 7, 11]. In our patient, both feet were painful at the calcaneocuboid joint and the first metatarsophalangeal joint. The former was probably the result of an overload at the calcaneocuboid joint, whereas the latter constituted an incipient hallux rigidus, probably related to the metatarsus primus elevatus. The patient's relatives were asymptomatic.

We found normal subtalar motion on physical examination and presumed it could be explained by characteristics observed on the radiographs; the broad, anterior side of the fused navicular was more uniformly and smoothly convex than usual; the normal mildly faceted appearance was absent. This appearance suggested the navicular-cuneiform joints had acquired some of the functions of the normal talonavicular joint. This also could explain the normal subtalar motion of both feet, because a fused talonavicular joint should have almost complete restriction of inversion-eversion movement.

Talonavicular fusion may have increased the mechanical load at the calcaneocuboid joint. Distraction calcaneocuboid joint arthrodesis was performed to relieve pain and to improve alignment of both feet (hyperpronation). First metatarsal plantar flexion osteotomy also was performed to correct the metatarsus elevatus and improve metatarsophalangeal joint pain.

We observed talonavicular synostosis associated with other unreported orthopaedic anomalies. Although this condition is less likely to be important clinically than other tarsal fusions, it sometimes can be painful enough for the patient to undergo surgery.

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