Case Report

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Treatment of isolated talonavicular coalition: Case report and literature review

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Abstract

Tarsal coalition refers to an abnormal fibrous, cartilaginous, or bony connection that develops between two or more tarsal bones. Talocalcaneal coalition and calcaneonavicular coalition account for more than 90% of all cases of tarsal coalition. Coalition exists early at birth, but bony connection usually develops during the patient's late growth period. Isolated cases of talonavicular coalition have rarely been reported. We herein report a case involving an 11-year-old patient with an isolated talonavicular coalition from a soft tissue to bony connection who was treated with arthroscopy for ankle arthritis. To our knowledge, this is the first case in which the whole formation of the talonavicular coalition was observed with a series of radiographic and magnetic resonance imaging examinations. The pain caused by the talonavicular coalition was managed by nonoperative treatment, while the ankle pain caused by the arthritis was relieved after ankle arthroscopy. At 6 years postoperatively, the patient remained pain-free while walking for 30 minutes and was satisfied with the operative outcome. Continuous follow-up confirmed that after the formation of talonavicular coalition, the coalition can continue to progress, forming bony talocalcaneal coalition and calcaneocuboid coalition.

Keywords

Talonavicular joint, coalition, treatment, arthroscopy, arthritis, radiography, magnetic resonance imaging, deformity

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Introduction

Coalitions can be categorized as abnormal syndesmosis, synchondrosis, or synostosis, and the most common types of tarsal coalition occur in the talocalcaneal joint and calcaneonavicular joint.[1–3] The reported prevalence of tarsal coalition ranges from 1% to 2%, but Nalaboff and Schweitzer[4] found a high prevalence of 11.5% in their prospective study. In another study, the prevalence was even higher at 13%.[5] About 50% of tarsal coalitions are bilateral.[1,6]

The most common type of tarsal coalition is talonavicular coalition, and the second most common type is calcaneonavicular coalition; coalition in other foot and ankle joints have also been reported.[7] The pathogenesis of tarsal coalition remains unclear and quite controversial.

There are no detailed reports on how bony tarsal coalition gradually develops after birth. Additionally, although many reports on the treatment of tarsal coalition have been published, few have described how to treat coalition with concomitant ankle arthritis. In this study, we followed a case involving the successful treatment of tarsal coalition formed by spontaneous connection of the talonavicular joint for unknown reasons.

Case report

An 11-year-old girl was admitted to our hospital on 7 August 2012 with a >2-year history of swelling and pain in the left ankle. The patient denied a history of trauma. Unexplained pain occurred in the left ankle after walking and was relieved after resting. She had no fever or other joint discomfort.

Before presentation at our hospital, a venous blood test at a local hospital showed an erythrocyte sedimentation rate (ESR) of 33 mm/hour, white blood cell (WBC) count of 11.1×10^9/L, and negative titers for anti-streptolysin O, rheumatoid factor, and tuberculous antibody. The first radiograph of the left foot taken after symptom onset showed no obvious abnormality. The patient's symptoms did not improve after oral administration of nonsteroidal pain relievers and conservative treatment. She was then transferred to another local hospital. Blood re-examination showed that her ESR had returned to a normal value of 11 mm/hour and her WBC count had returned to 6×10^9/L. Common indicators of connective tissue disease, such as human leukocyte antigen B27, anti-keratin antibody, anti-cyclic citrullinated peptide antibody, and autoantibody, were all negative. Radiographic re-examination showed slight narrowing of the left talonavicular joint space; the lower limb alignment was basically normal. Magnetic resonance imaging (MRI) examination revealed obvious manifestations of arthritis in the left talonavicular joint, obvious edema in the talus and navicular bone, and effusion in the ankle and subtalar joints with hyperplasia of the synovial tissue between them (Figures 1–8). The local pain was not relieved after conservative treatment in the second local

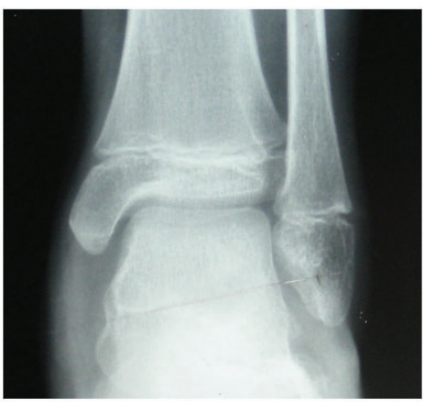

Figure 1. First ankle radiograph 6 months after symptom onset suggesting no obvious abnormalities in the bony structure (anteroposterior view).

Figure 2. First ankle radiograph 6 months after symptom onset suggesting no obvious abnormalities in the bony structure (lateral view).

Figure 3. Ankle radiograph 8 months after symptom onset suggesting mild talonavicular arthritis (lateral view).

hospital; thus, the patient was transferred to our hospital for further treatment.

Physical examination at our hospital revealed left ankle swelling and tenderness, limitation of eversion and inversion of the ankle, no obvious limitation of ankle dorsiflexion or plantar flexion, and no severe deformity in the lower limb alignment. Venous blood tests showed a normal ESR

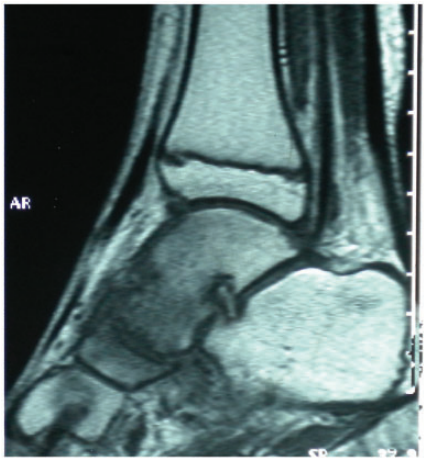

Figure 4. Ankle magnetic resonance image 8 months after symptom onset suggesting talonavicular arthritis (sagittal T1 sequence).

Figure 5. Ankle magnetic resonance image 8 months after symptom onset suggesting talonavicular arthritis and bone edema (sagittal STIR sequence).

and WBC count and negative antistreptolysin O, rheumatoid factor, antikeratin antibody, anti-cyclic citrullinated peptide antibody, human leukocyte antigen B27, autoantibody spectrum, and tuberculous antibody. A chest radiograph showed

Figure 6. Ankle radiograph 12 months after symptom onset suggesting no formation of talonavicular coalition (lateral view).

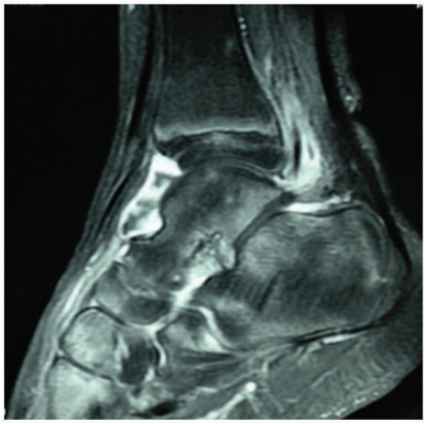

Figure 7. Ankle magnetic resonance image 12 months after symptom onset suggesting ankle arthritis (sagittal STIR sequence).

no abnormality, while a left foot radiograph showed bony connection of the talonavicular joint, forming a tarsal coalition. MRI examination confirmed the bony connection of the talonavicular joint and revealed ankle joint effusion with low signal intensity. The effusion was obtained for bacterial culture and tuberculosis polymerase chain reaction; the results were negative, and bacterial infection and tuberculosis were therefore ruled out. The patient's condition was becoming slowly and progressively aggravated, and the talonavicular joint had spontaneously connected. Moreover, the subtalar and

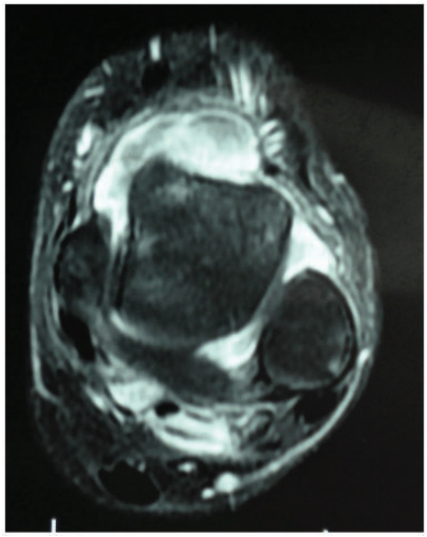

Figure 8. Ankle magnetic resonance image 12 months after symptom onset suggesting ankle arthritis and massive hyperplasia of synovial tissue (axial STIR sequence).

ankle joints began to be involved, and the MRI results suggested that the lesion was located within the ankle joint, resulting in specific manifestations of ankle joint effusion and a space-occupying lesion in the joint cavity. After rest and oral analgesics, the patient's condition continued to progress. We therefore planned to perform pathological examination to confirm the diagnosis and debridement of the hyperplastic synovial tissue to slow the progression of the disease. After a thorough discussion with the patient and her parents, we performed surgery under nerve block anesthesia on 13 August 2012.

The surgeon followed routine arthroscopic procedures[8] and conducted an arthroscopic exploration. Hyperemia of the synovial membrane and nodular hyperplasia could be seen under arthroscopy; no obvious damage was observed in the articular cartilage. A specimen of the synovial membrane was taken for pathological examination. A planer was then used to remove the entire intra-articular synovial

lesion. After the surgery, the patient was given a plaster cast for immobilization and rested for 1 week. When the stitches were taken out 2 weeks postoperatively, weight-bearing walking was conducted under the protection of walking boots. An analgesic and traditional Chinese medicine were taken orally. Regular follow-ups were conducted. The patient's pain during walking was significantly relieved after 4 months. The patient was followed for 6 years, and her visual analog scale score decreased from 8 to 1. There was no obvious flatfoot deformity in the left ankle when standing (Figures 9–19, Video 1). However, the gait of the left side was abnormal compared with the right side while walking. We could not rule out the need for further treatment to alleviate the abnormal gait and potentially worsening pain.

This study was conducted in accordance with the principles of good clinical practice and the Declaration of Helsinki and was approved by the Southwest Hospital Ethics Review Committee (No. 2012-8254940). The patient provided written informed consent for publication.

Figure 9. Ankle radiograph 24 months after symptom onset suggesting formation of talonavicular coalition (lateral view).

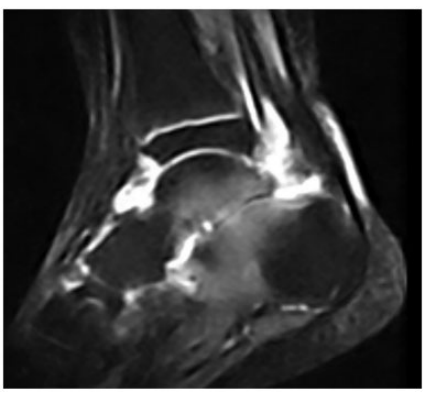

Figure 10. Ankle magnetic resonance image 24 months after symptom onset suggesting completed talonavicular coalition and arthritis located at the ankle and subtalar joint (sagittal STIR sequence).

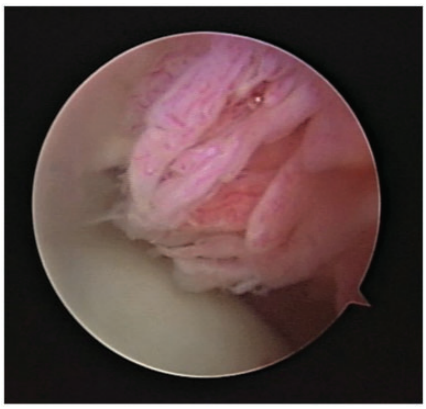

Figure 11. Arthroscopic exploration revealing synovial congestion and hyperplasia.

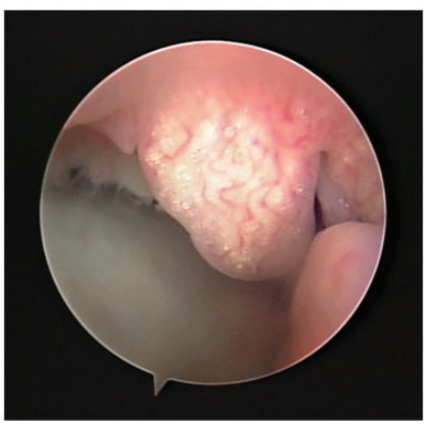

Figure 12. Some of the synovial hyperplasia is nodular with visible pannus.

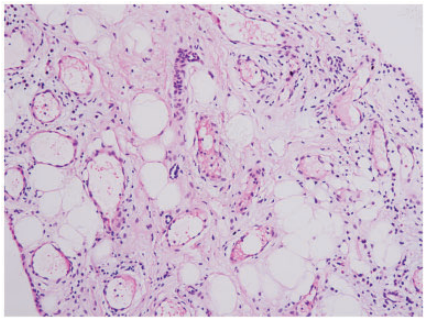

Figure 13. Pathological examination of the synovial tissue showing formation of inflammatory granulation tissue (hematoxylin–eosin staining, ×200).

Figure 14. Left ankle swelling was relieved 2 weeks after arthroscopy.

Discussion

The cause of tarsal coalition is unclear

Tarsal coalition reflects abnormalities in one or more tarsal joints.[9] It usually occurs between two adjacent tarsal bones and can be classified as talocalcaneal coalition, calcaneonavicular coalition, calcaneocuboid coalition, talonavicular coalition, and other types of coalition. The most common types are talocalcaneal and calcaneonavicular coalitions; in contrast, calcaneocuboid and talonavicular coalitions are much more rare. The cause of tarsal coalition remains unclear, but previous studies have promoted hypotheses regarding congenital and acquired origins.[10]

Figure 15. Left ankle swelling continued to be relieved 4 months after arthroscopy.

Figure 16. No obvious swelling was present in the left ankle 5 years after arthroscopy.

Figure 17. The medial arch of the left foot was still present 5 years after arthroscopy.

Figure 18. Ankle radiograph 8 years after symptom onset suggesting talonavicular, subtalar, and calcaneocuboid coalition; however, the arch of the foot was still good (lateral view).

Although tarsal coalition exists at birth, synostosis usually occurs later in the patient's growth period. Tarsal coalition is a malformation because one or more tarsal joints are usually lacking; it is also a deformity because hindfoot valgus or varus often develops secondarily.[11] Coalition may not be symptomatic at first; it gradually becomes so as the coalition ossifies. In most cases of calcaneonavicular coalition, this occurs from 8 to 12 years of age, whereas patients with talocalcaneal coalition develop symptoms around 12 to 16 years

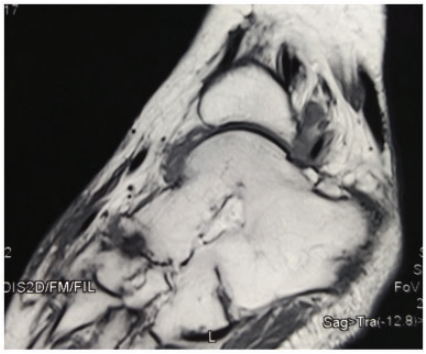

Figure 19. Ankle magnetic resonance image 8 years after symptom onset suggesting talonavicular, subtalar, and calcaneocuboid coalition with degeneration of the cuneonavicular joint; however, the ankle joint space remained normal (sagittal T1 sequence).

of age.[12] Congenital tarsal coalition is autosomal dominant and the penetrance is quite strong; any type of coalition can be inherited.[7]

In addition to the patient's genetic factors, other contributing factors for the formation of tarsal coalition include juvenile rheumatoid arthritis, trauma, and infection.[2] However, these factors are difficult to confirm through clinical examination and are usually only suspected. In the present case, we conducted a complete and detailed follow-up of the patient's talonavicular coalition. A series of radiographs and MRI examinations confirmed the tarsal coalition, and the talonavicular joint space was completely filled. The course of disease was about 2 years, and we speculated that it was related to local juvenile rheumatoid arthritis. In addition to a transient increase in the ESR and WBC count, all venous blood indicators of rheumatoid arthritis were negative. During the patient's disease course, the talonavicular joint was first moth-eaten, the joint surface became uneven, and then osseous tissue gradually formed. MRI suggested that edema had developed in the osseous tissues on both

sides of the talonavicular joint. If this was juvenile rheumatoid arthritis, the abnormal signals could also affect the subtalar and ankle joints. Effusion and hyperplasia of synovial tissue could be seen in the ankle joint. These inflammatory reactions can cause pain in more extensive areas. Because the disease might progress further, it is important to maintain the foot of the affect limb in a functional position.

Status of tarsal coalition treatment and value of arthroscopic application in this case

Coalitions are often asymptomatic. In such cases, a low level of arthritis is expected and treatment should be personalized. Treatment options for coalitions usually start with nonoperative methods for at least 6 months and include rest from intense exercises, physical therapy to stretch the Achilles tendon, treatment for sprained ankle, anti-inflammatory medications, short-term immobilization for 3 to 4 weeks under the protection of walking boots, and orthotics to support the arch and medial heel. If these nonoperative methods all fail, surgery can be considered. Arthrodesis should be performed only as a last option after several treatments have failed.[1,7]

If the talonavicular joint is in a nonfunctional position, it may need to be surgically adjusted to a functional position. Three-dimensionally printed personalized lesion models will help to improve the surgical plan.[13] If the bony connection is in a functional position, the symptoms of the talonavicular joint are usually mild while the adjacent joints develop arthritis. When the MRI results show significant hyperplasia of the synovial tissue and lesions within the joint cavity, arthroscopy will have significant therapeutic value in reducing the inflammatory reaction of the ankle joint. Arthroscopic techniques have been rapidly developed in recent years with respect to minimal trauma and complete removal of synovial tissue from the joint cavity.[8,14,15] In the present case, we observed obvious hyperplasia of the synovial tissue in the ankle joint as well as pannus formation under arthroscopy. However, although arthroscopy can completely debride the synovial tissue and reduce inflammation, it has a learning curve and requires a high level of technical skill to ensure its efficacy.

In this study, we followed the complete process of talonavicular coalition formation. The disease course was about 2 years, and juvenile rheumatoid arthritis may have been an important causative factor. Continuous follow-up confirmed that after formation of the talonavicular coalition, such lesions can continue to progress, forming bony talocalcaneal coalition and calcaneocuboid coalition. Arthroscopic debridement has a certain therapeutic value when coalition is combined with ankle arthritis.

Declaration of conflicting interest

The authors declare that there is no conflict of interest.

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