



## Realignment Calcaneal Osteotomy for Bilateral Complete Talocalcaneal Synostosis: A Case Report

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### ABSTRACT

Talocalcaneal synostosis is a congenital failure of the segmentation between tarsal bones. It may be very difficult to differentiate from talocalcaneal coalition, known as the most common tarsal coalition, especially in cases with a large bone bridge. Complete talocalcaneal synostosis is very rare, and there are few references in the literature about the clinical outcomes and operative methods for symptomatic synostosis. We report a case of a 15-year-old female with bilateral complete talocalcaneal synostosis and heel varus deformity who has experienced good clinical results after lateral sliding calcaneal osteotomy for hindfoot realignment.

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Tarsal coalition is believed to result from a failure of embryonic mesenchymal differentiation and segmentation (1). The bony bridge between tarsal bones can involve a partial portion or the entirety of the joint space (2,3). The anatomic location of coalition and the extent of involvement are important in determining the symptoms and appropriate operative treatment method. Tarsal coalition usually becomes symptomatic around the ages of 12 to 16, which correlates with the time that the coalition begins to ossify (4). These symptoms include limitation of subtalar joint motion, discomfort at walking, ankle pain from subfibular impingement, peroneal spastic flatfoot, and pes planovalgus deformity (1). Talocalcaneal and calcaneonavicular coalitions are bilateral ~50% to 60% of the time (5,6). Tarsal coalitions are usually known to be associated with planovalgus deformity, but cavovarus deformity can occur very rarely. Although a few cases have been described in the literature (7–9), the pathologic mechanism leading to cavovarus deformity in the presence of tarsal coalitions remains unclear. Stuecker and Bennett (7) reported 3 patients who presented with cavovarus deformities without underlying neurologic abnormalities: 1 with a calcaneonavicular coalition and 2 with talocalcaneal coalitions. Merchant et al (8) also described a case of heel varus deformity with massive tarsal coalitions in multiple synostosis syndrome. However, our inspection of the current published data failed to identify the exact percentage of hindfoot varus deformity in tarsal coalitions.

We describe a case with symptomatic talocalcaneal synostosis involving the entire subtalar joint. A literature review did not yield

reports about definitive surgical methods after conservative treatment fails for this condition. We placed the main focus of surgical treatment on the correction of hindfoot varus malalignment, as opposed to the restoration of subtalar joint motion. Given the previously described background, we report a case of a patient with bilateral complete talocalcaneal synostosis and unilateral heel varus deformity who has experienced satisfactory clinical outcomes and improvement of gait function through hindfoot realignment calcaneal osteotomy.

### Case Report

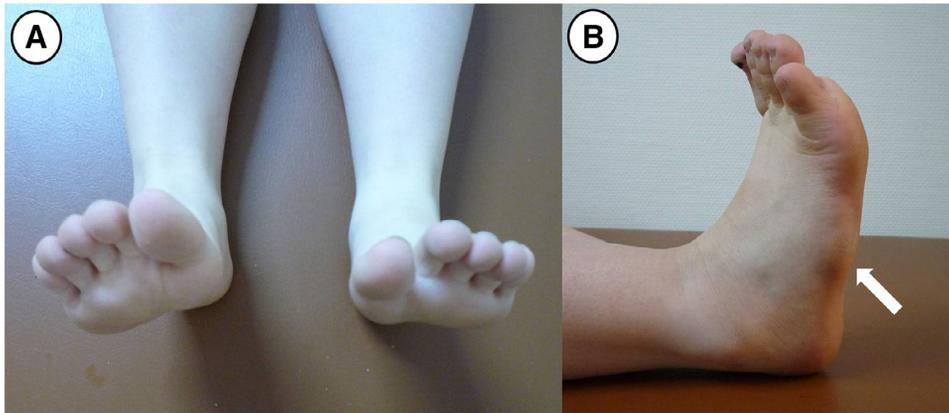
A 15-year-old female patient complained of pain at the lateral aspect of the right foot, discomfort with walking on uneven ground, and difficulty in performing strenuous activities for 2 years before presentation. She had no history of ankle or foot injury. On physical examination, both subtalar joints had severely restricted range of motion, and heel varus deformity of the right foot was observed. Although plantarflexion of the ankle joint was normal, dorsiflexion was limited to the neutral position, and passive dorsiflexion induced discomfort secondary to heel cord tightness. Painful callus with focal tenderness was observed at the lateral border of the right foot (Fig. 1). No significant cavus or pes planus deformity was observed. Although the circumference of the 2 calves was similar, there was an approximately 1-cm limb length discrepancy as determined by the spine–malleolar distance. On measurement based on computed tomography (CT) scan, the right lower limb was slightly shorter than the left. There was no specific trauma or disease on medical history. However, she complained of a deficiency of flexibility in both ankles when she was exercising, running, or walking on uneven ground. The patient did not note pain with daily activities, even though she recognized the limited motion of both subtalar joints. On radiographic examination, bilateral complete talocalcaneal synostosis was

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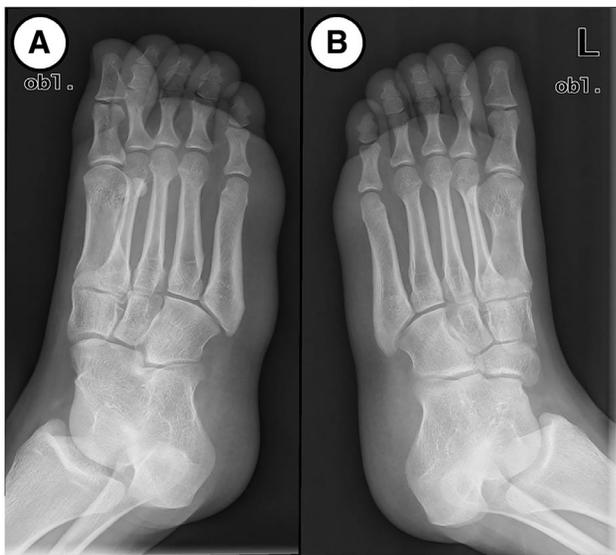
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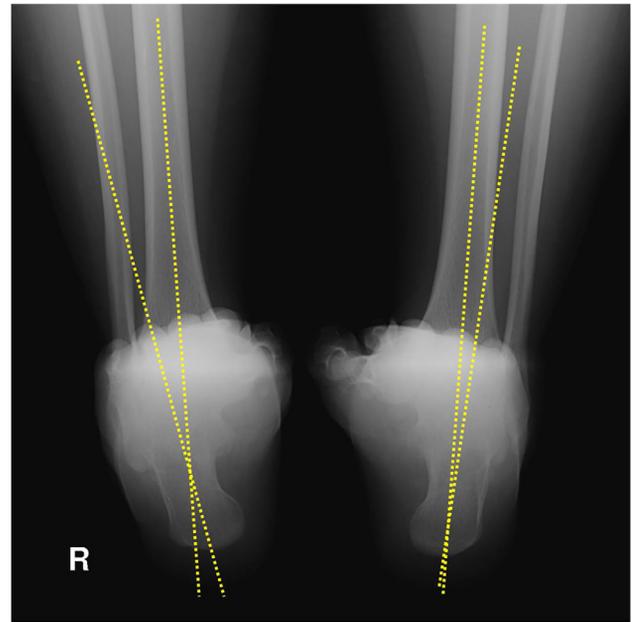


**Fig. 1.** (A) Preoperative photograph shows mild limb length discrepancy and varus malalignment observed in the plantar surface of the right foot. (B) Preoperative photograph shows limitation of the ankle dorsiflexion and a painful callosity (arrow) at the lateral border of the right foot.



**Fig. 2.** Preoperative oblique radiographs show bilateral talocalcaneal synostosis. (A) Right. (B) Left.

found (Figs. 2 and 3). The hindfoot alignment view demonstrated a calcaneal varus deformity of the right foot compared with the contralateral side (Fig. 4). A CT scan of the right and left feet revealed that the coalition between the talus and calcaneus demonstrated complete continuity of cortical and cancellous bone through the entire subtalar joint (Figs. 5 and 6).



**Fig. 4.** Preoperative hindfoot alignment view shows heel varus deformity of the right foot compared with the contralateral side.

After a trial of a lateral heel-wedge insole for 3 months without improvement of symptoms, we proceeded with surgical treatment to equalize pressure distribution through hindfoot realignment of the right foot. Under spinal anesthesia, the patient was placed in the prone



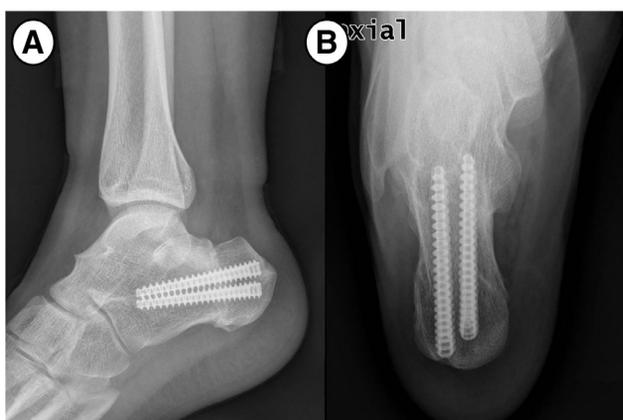
**Fig. 3.** Preoperative standing lateral radiographs show bilateral complete talocalcaneal synostosis without the pes planus deformity. (A) Right. (B) Left.



**Fig. 5.** Preoperative coronal (A) and sagittal (B) images of CT scan on the right ankle show complete talocalcaneal synostosis.

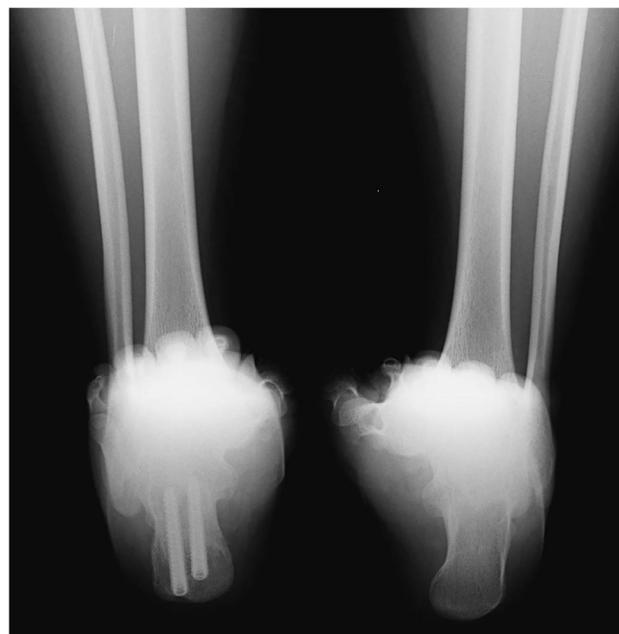


**Fig. 6.** Preoperative coronal (A) and sagittal (B) images of CT scan on the left ankle show complete talocalcaneal synostosis.



**Fig. 7.** Follow-up lateral (A) and axial (B) radiographs 3 years postoperatively demonstrate union of osteotomy site after the lateral sliding calcaneal osteotomy and stable fixation using headless compression screws.

position with a thigh tourniquet. First, Achilles tendon lengthening using the triple hemisection technique was performed to resolve the heel cord tightness. Next, a 3-cm oblique incision was made in line with the peroneal tendons to expose the lateral wall of the calcaneus. An oblique calcaneal osteotomy with a 3-mm closing wedge through a resection of the lateral wall was performed, and the tuberosity portion was displaced laterally. We used a calcaneus long axial view intraoperatively to determine the appropriate degree of lateral displacement and tried to obtain  $\sim 0^\circ$  to  $\sim 5^\circ$  of hindfoot valgus alignment. In addition, we also obtained  $\sim 1$  cm of inferior translation to reduce the limb length discrepancy. After the correction of the hindfoot, varus malalignment



**Fig. 8.** Follow-up radiograph 3 years postoperatively demonstrates restoration of the hindfoot alignment.

was checked under intraoperative fluoroscopy, and the osteotomy site was fixed with 2 headless compression screws (Fig. 7).

A short leg cast and non-weightbearing ambulation with a crutch were maintained for 4 weeks postoperatively. Thereafter, gentle range-of-motion exercises and partial weightbearing ambulation were encouraged. Full weightbearing gait was permitted at 8 weeks postoperatively. Although the limitation of subtalar joint motion was unchanged, the patient's painful callus and heel varus deformity were resolved after surgery (Fig. 8). The patient showed satisfactory clinical outcomes and functional activities (without discomfort while walking) on the latest follow-up visit 8 years postoperatively (Fig. 9).

## Discussion

Synostosis results when 2 bones are united and motion is restricted completely. This cross-union is most commonly seen as a rare complication of forearm fractures in children or from distal tibiofibular syndesmotic injury. The proper use of the terms synostosis or coalition is still unclear to describe the case in this study. We propose that the use of synostosis is reasonable because talus and calcaneus had a complete continuity of cortical and cancellous bone through the entire articular surface of the subtalar joint. In addition, the case in this study demonstrated hindfoot varus malalignment, in contrast with most cases of conventional tarsal coalitions. We have hypothesized that this case may have been a congenital difference (such as a failure of differentiation of embryonic mesenchymal tissue), because there was no specific trauma or disease on review of the medical history of the patient. However, we could not reach a clear conclusion regarding pathophysiology owing to a deficiency of prior medical records or radiographs in consecutive long-term follow-up visits from childhood.

The incidence of tarsal coalition has been reported from 1% to as high as 6% among the general population (1–3). There are several proposed theories related to the etiology of tarsal coalitions. Of these, the most widely accepted is LeBouq's theory of failure of differentiation of embryonic mesenchymal tissue, believed to be a heritable autosomal dominant defect or secondary to an insult sustained in the first

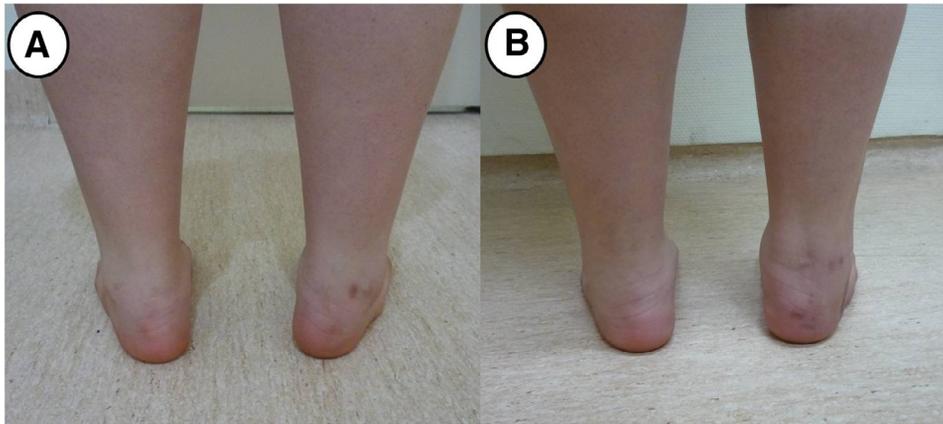


Fig. 9. (A) Preoperative photograph shows heel varus malalignment of the right foot. (B) Follow-up photograph 3 years postoperatively shows restoration of the hindfoot alignment.

trimester of pregnancy (2). However, there have been reports that coalitions occurred from ossification of accessory bones into the adjacent tarsal bones and were also acquired from arthritis, infection, neoplasm, or trauma (2,6,10).

Most tarsal coalitions become symptomatic in the early adolescence period, with micromotion noted at the articulation (4). The talocalcaneal bridge ossifies either completely or incompletely when an individual is between 12 and 16 years old, which is later than the ossification of the calcaneonavicular bar and usually is diagnosed in older adolescents or adults. We believe that talocalcaneal coalition with complete bony bridge is extremely rare. However, there is the possibility that cases with complete bony bridge are less symptomatic than cases with partial bony bridge or cartilaginous bar. If the subtalar joint is fused completely by a bony bridge, the pain may be relatively mild and patients may not seek treatment. A patient with total bilateral tarsal coalition (synostosis of all the tarsals) reported by Mettu et al (11) had reached 35 years of age without significant disability. Furthermore, the patient with congenital subtalar fusion may feel relatively less uncomfortable without insidious limitation of subtalar joint motion.

Depending on the extent of coalition and the status of adjacent tarsal joints, surgical treatments for talocalcaneal coalition consist of resection and interpositional fat or muscle graft, subtalar arthrodesis, triple arthrodesis, subtalar arthroereisis, and calcaneal lengthening osteotomy (12). To the best of our knowledge, a case with bilateral talocalcaneal synostosis involving the entire subtalar joint has not been previously presented in the literature. We attempted to find an optimal surgical method for this challenging problem after conservative treatment had failed. We placed the main focus of surgical treatment on the correction of hindfoot varus malalignment, as opposed to the restoration of subtalar joint motion. Symptoms from the hindfoot malalignment can be present and be misinterpreted as being purely from the coalition. This may lead to isolated resection of the coalition without correction of the symptomatic hindfoot deformity. The resection of the coalition without surgical treatment of the symptomatic flatfoot deformity has been reported to result in increased deformity and failure of the clinical result (13). In patients with large talocalcaneal coalitions associated with rigid hindfoot valgus deformity, Mosca and Bevan (12) have reported that calcaneal lengthening osteotomy and Achilles tendon lengthening without coalition resection is effective for correcting deformity and relieving pain in rigid flatfoot.

Strong correlation has been found between the size of the coalition and the clinical outcome (14). Wilde et al (13) reported that resection of coalition is contraindicated in talocalcaneal coalitions

occupying greater than one third of the total articular surface of the subtalar joint. Hetsroni et al (15) reported that despite the favorable clinical outcome observed in tarsal coalition patients after bar resection, foot kinematics are not fully restored. This is expressed as severe restriction of subtalar motion observed during walking, and additional operative procedures or specific postoperative rehabilitation programs may be necessary for the improvement of foot kinematics (15).

The other important feature of this case is the associated hindfoot deformity. Most patients with tarsal coalition have a fixed hindfoot valgus (rigid planovalgus deformity) of varying severity (16). Less well known is the association of hindfoot varus with tarsal coalition. In tarsal coalition, peroneal spasm can occur to effectively reduce the pressure on the subtalar joint, which acts as the cause of pain. This can result in secondary adaptive shortening of the peroneus longus, soleus, and gastrocnemius muscles in response to heel valgus (17,18). Similarly, the hindfoot varus deformity of the patient in this case study may have resulted from an adaptation to the limitation of subtalar motion. However, we could not find any report that varus deformity of the hindfoot occurs after therapeutic subtalar arthrodesis in pediatric patients.

In conclusion, a 15-year-old female patient with bilateral complete talocalcaneal synostosis and unilateral heel varus deformity showed satisfactory clinical results and improvement of gait function after lateral sliding calcaneal osteotomy for hindfoot realignment.

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