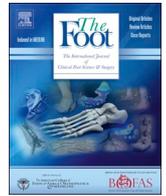




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Case Report

Tibial spastic varus foot caused by os calcaneus secundarius: A case report

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ABSTRACT

Peroneal spastic flatfoot caused by tarsal coalition is well known; however, tibial spastic varus foot is a rare clinical entity also caused by tarsal coalition in most cases. The os calcaneus secundarius is a rare accessory bone between the anterior process of the calcaneus and the navicular bone. The case of a 29-year-old woman with tibial spastic varus foot caused by os calcaneus secundarius is presented. Operative excision of the os calcaneus secundarius completely resolved the varus deformity. This is the first case report involving tibial spastic varus foot caused by os calcaneus secundarius.

1. Introduction

It is well known that tarsal coalition causes the peroneal spastic flatfoot (PSFF) deformity; in contrast, tibial spastic varus foot (TSVF) deformity is rare, and most cases are caused by tarsal coalition [1–10]. The os calcaneus secundarius (CS) is an accessory bone of the foot that is incidentally found on radiography or computed tomography (CT) during the assessment of foot or ankle pain [11]. To our knowledge, TSVF caused by CS is unreported in the literature. In this paper, a case of a 29-year-old woman with TSVF caused by CS that was completely resolved by surgical resection. The patient provided written informed consent for the publication of her case.

2. Case report

A 29-year-old female chef presented to our outpatient clinic with a 3-month history of right foot pain and varus deformity of her right ankle. Fifteen months prior, she suffered an eversion sprain of the right ankle when she slipped into a gutter while walking. The right ankle pain had decreased gradually since the injury; however, a varus deformity developed 2 months before she presented to our clinic, especially after work, with gradually increasing frequency. She was obese with a height of 166 cm and body weight of 92 kg. The patient's neurological findings were normal. A physical examination revealed that her right ankle had a varus deformity and the tibialis anterior tendon was tense (Fig. 1). The anterior aspect of the sinus tarsi was tender, and a small bony prominence was palpable anterior to the sinus tarsi. No

swelling or erythema of the affected foot was noted. The range of motion of the right ankle was normal. The varus deformity was manually corrected but without passive valgus force. Plain radiography revealed the anterior calcaneal bar extending toward the navicular bone (Fig. 2a). Reconstructed three-dimensional CT (3D-CT) showed a 20 mm × 15 mm × 8 mm well-demarcated triangular ossicle in the calcaneonavicular joint (Fig. 2b). No extra ossicle was found in the contralateral foot. Multiple cysts with sclerotic changes were observed in the anterior process (Fig. 2c). Magnetic resonance imaging (MRI) demonstrated abnormal intensity in the anterior process and calcaneus just below the anterior process (Fig. 2d). A lidocaine injection into the sinus tarsi transiently relieved the foot pain but did not correct the varus deformity. A possible diagnosis was TSVF caused by CS or pseudoarthrosis of the anterosuperior process of the calcaneus with or without calcaneonavicular coalition. Because conservative therapy with nonsteroidal anti-inflammatory drugs for 3 weeks failed to relieve the pain or correct the ankle deformity, surgical resection of the extra ossicle was performed. The ossicle was not connected to the adjacent bones and was easily excised. Pathological examination revealed that all surfaces of the excised ossicle were surrounded by normal cartilage; no osseous or fibrous union was observed (Fig. 3a and b). Finally, CS instead of pseudoarthrosis of the anterior process of the calcaneus was the likely diagnosis. A short leg cast was applied with neutral position of the ankle for 2 weeks. A postoperative reconstructed 3D-CT showed that the CS was completely resected (Fig. 4). Two years after the operation, the right foot pain and varus deformity were completely resolved with no limitation of right ankle motion, and the patient had no

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Fig. 1. Preoperative photograph of the patient's foot showing a right ankle varus deformity with tense tibialis anterior muscle and tendon (arrowheads).

complaints about the affected ankle.

3. Discussion

CS is an accessory ossicle of the foot that is located at the gap between the anteromedial aspect of the calcaneus, proximal aspect of the cuboid and navicular bone, and head of the talus [11]. CS probably has a congenital or genetic origin, resulting in an accessory center of ossification [12,13]. Because its prevalence is rare, 3.4% in 1367 specimens according to Mann [12], CS has not been widely recognized by orthopedic surgeons compared to other prevalent accessory bones of the foot such as the accessory navicular bone, os peroneum, os trigonum, or os subfibulare [11]. Although several cases of symptomatic CS have been reported [14–18], CS is asymptomatic in most cases and usually found incidentally by radiographic assessment, especially in the examination of the cause of ankle pain after a sprain. Therefore, CS mimics fracture of the anterior process of the calcaneus or a calcaneonavicular coalition, as in our case [14,16,19,20]. The pathological examination of the specimens revealed no fibrous or callus tissues but only normal cartilage on all surfaces of the ossicle, leading to the final diagnosis of CS.

PSFF is a well-known foot deformity following tarsal coalition; on the contrary, TSVF caused by pathology in the subtalar joint is rare. Only 17 cases have been reported to date [1–10]. Of them, 15 were accompanied by calcaneonavicular or talocalcaneal coalition, one with osteoid osteoma of the calcaneus [8], and one with accessory anterolateral talar facet impingement [5]. TSVF caused by CS has not been reported to date. Although the mechanism of TSVF caused by subtalar lesions has not been fully elucidated, the “escape phenomenon” from foot pain is the most probable cause of TSVF [3,4,7]. To prevent foot pain, tibial muscle spasms can develop. In our case, an axial load to the heel and eversion stress to the ankle were likely the precedent injury

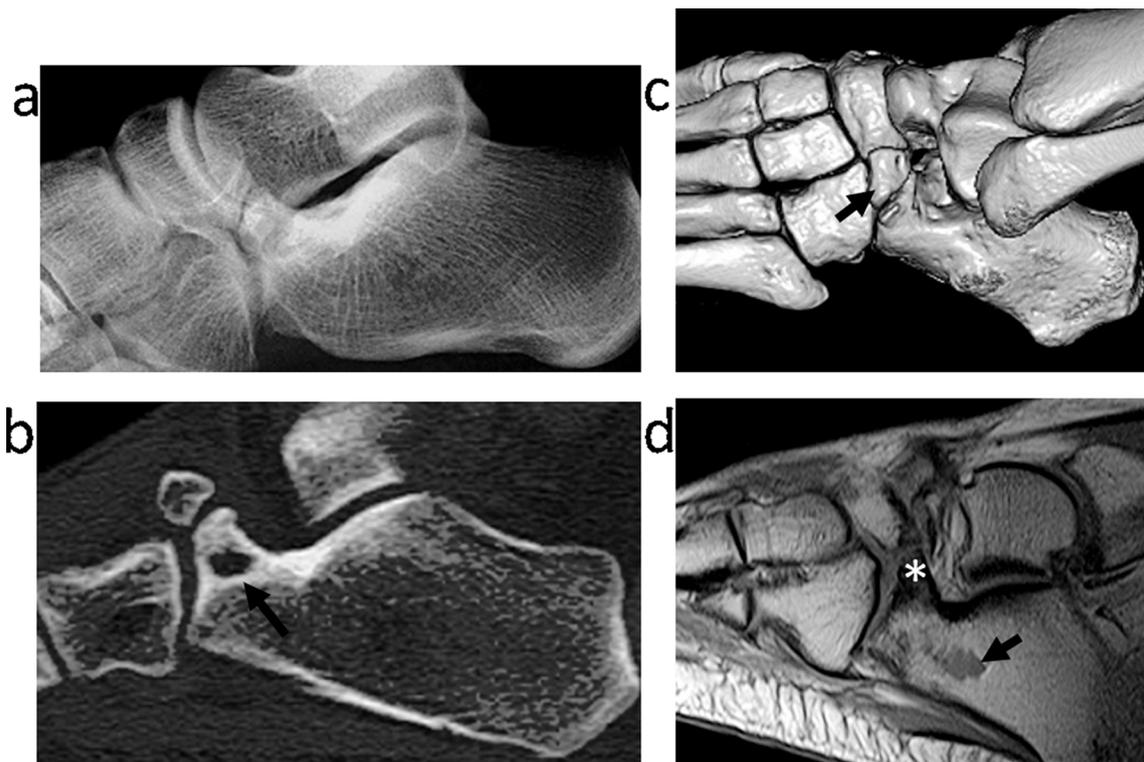


Fig. 2. Preoperative radiography, computed tomography, and magnetic resonance imaging findings. (a) Preoperative lateral radiography showing the calcaneonavicular bar. (b) Reconstructed three-dimensional computed tomography image revealing a triangular extra ossicle (black arrow) between the calcaneus and navicular bones. (c) Sagittal image on computed tomography showing cystic and sclerotic changes to the anterior process of the calcaneus bone (black arrow). (d) T1-weighted magnetic resonance image showing a low-intensity signal of the anterior process of the calcaneus bone (white asterisk) and just below the calcaneus bone (black arrow).

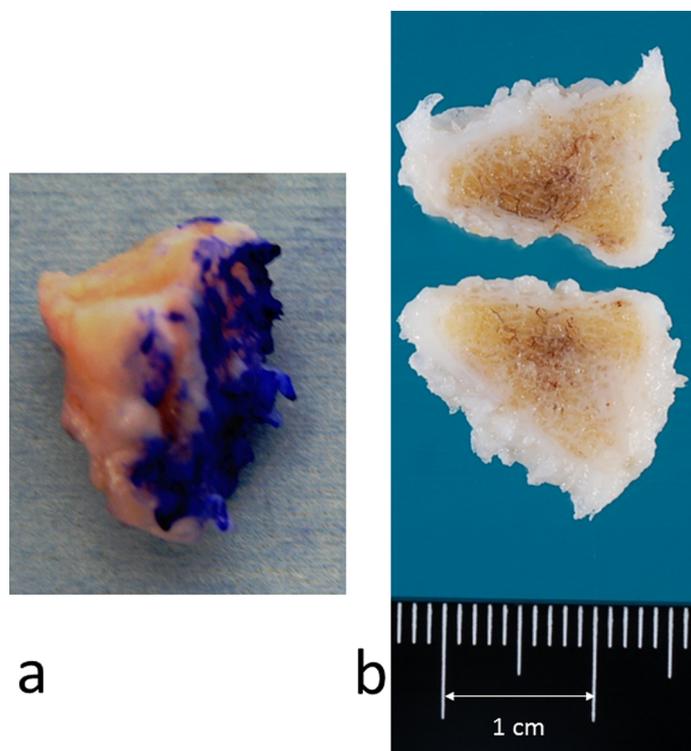


Fig. 3. (a) Resected ossicle is shown. The surface facing the navicular bone is painted. The excised ossicle is triangular in shape. (b) The parting plane indicates that all surfaces are covered with intact articular cartilage, leading to the diagnosis of os calcaneus secundarius.

mechanisms as suggested by the MRI results, which showed signal intensity changes in the anterolateral part of the calcaneus bone. Moreover, daily repetitive stress to the lateral subtalar joint due to obesity might be responsible for the osteoarthritic change seen on CT and MRI. Since the subtalar motion was probably restricted due to CS having a similar biomechanism to calcaneonavicular coalition, axial and eversion stress could not be neutralized by the subtalar joint but could be directed to impact the calcaneus because of CS, an inclusion in the subtalar joint. Ceroni et al. also reported a 13-year-old boy with symptomatic CS with limited subtalar joint presenting a similar biomechanical cause to calcaneonavicular coalition [15]. CS resection is important to improving subtalar motion and decompressing impact stress to the anterolateral part of the calcaneus during weightbearing.

It is controversial whether symptomatic CS should be resected

[14,16]. Several case reports indicated that surgery should be performed only when conservative treatments, including anti-inflammatory drugs, avoiding weightbearing, and casting, have failed. Thus far, symptomatic CS resection has shown good clinical results [15,17,18]. Because our patient had TSVF and right ankle pain for a relatively longer period (3 months) and a lidocaine injection into the sinus tarsi was transiently effective, surgery instead of casting was selected. However, acquired varus foot deformities such as functional or psychogenic dystonia, in which surgery might be considered highly inappropriate, should be differentiated [21]. Our patient had no psychological disorders, family or work concerns, or neurological abnormalities. MRI findings and transient improvement with lidocaine injected into the lesion excluded the possibility of functional or psychogenic dystonia.

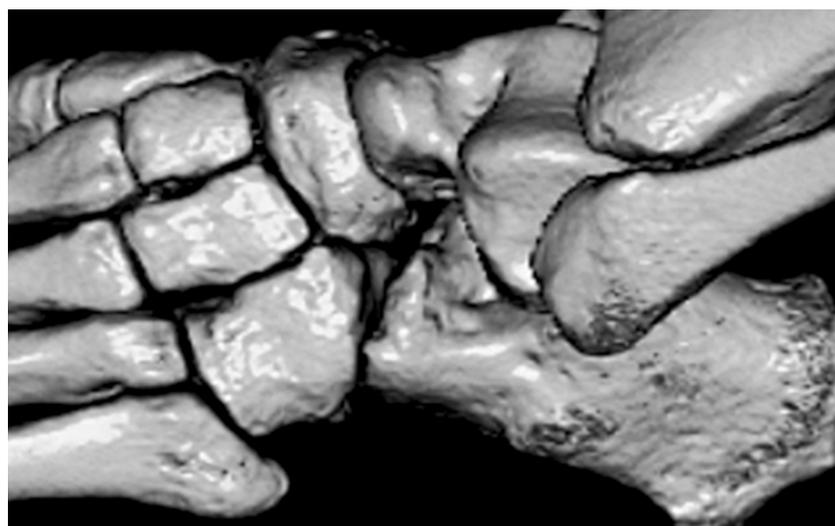


Fig. 4. Postoperative reconstructed computed tomography image showing a completely resected os calcaneus secundarius.

In conclusion, in this case, TSVF caused by CS, which has not been reported to date, was successfully treated by CS resection.

Conflicts of interest statement

The authors declare no conflict of interest.

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Consent

Informed consent has been taken from the patient for the publication of the clinical and radiographic data.

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