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Case Reports and Series

Complex Anatomic Abnormalities of the Lower Leg Muscles and Tendons Associated With Phocomelia: A Case Report

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ABSTRACT

Musculoskeletal anatomy is widely known to have components that stray from the norm in the form of variant muscle and tendon presence, absence, origin, insertion, and bifurcation. Although these variant muscles and tendons might be deemed incidental and insignificant findings by most, they can be important contributors to pathologic physiology or, more importantly, an option for effective treatment. In the present case report, we describe a patient with phocomelia and Müllerian abnormalities secondary to in utero thalidomide exposure. The patient had experienced recurrent bilateral foot pain accompanied by numbness, stiffness, swelling, and longstanding pes planus. These symptoms persisted despite conservative treatment with orthotics, steroids, and nonsteroidal anti-inflammatory drugs. Radiographic imaging showed dysmorphic and degenerative changes of the ankle and foot joints. Further investigation with magnetic resonance imaging revealed complex anatomic abnormalities, including the absence of the posterior tibialis and peroneus brevis, lateralization of the peroneus longus, and the presence of a variant anterior compartment muscle. The variant structure was likely a previously described anterior compartment variant, anterior fibulocalcaneus, and might have been a source of the recurrent pain. Also, the absence of the posterior tibialis might have caused the pes planus in the present patient, considering that posterior tibialis tendon dysfunction is the most common cause of acquired pes planus. Although thalidomide infrequently affects the lower extremities, its effects on growth and development were likely the cause of this rare array of anatomic abnormalities and resulting ankle and foot pathologic features.

Anatomic variations of the lower leg are quite common and can range from abnormal muscle origins, tendinous insertions, and bifurcations to the complete absence of structures such as the widely described variant of the lateral compartment, peroneus quartus. These variations are often asymptomatic and incidental findings. In some cases, however, certain anatomic variations can lead to chronic clinical problems. For example, lateral ankle pain, fibular tenosynovitis, and ankle instability have been attributed to peroneus quartus of the lateral compartment (1).

Thalidomide is a drug that was originally used in the 1950s by pregnant women as a sedative to prevent morning sickness. In 1961, thalidomide was found to be a teratogen that resulted in thousands of infant deaths, birth defects, and an unknown number of miscarriages (2). The most common abnormalities associated with thalidomide exposure involved phocomelia: congenital underdevelopment of the limbs with variable residual dysfunction. In phocomelia, the upper limbs are affected to a greater frequency and degree. Other commonly affected structures include the ears, eyes, reproductive organs, kidneys, gut, and nervous system. The exact mechanism has not been fully elucidated; however, the results from animal models have suggested correlation to inhibition of angiogenesis, reactive oxygen species-induced cell death, cell migration/adhesion inhibition via integrin underexpression, and direct effects on chondrogenesis (3).

Muscle and tendon abnormalities of the lower limb related to thalidomide exposure have not been as well documented as those for the upper limb. We describe a rare, complex, anatomic abnormality involving the various muscle compartments of a patient's lower leg and osseous structures of the ankle. These abnormalities were most likely caused by the patient's in utero exposure to thalidomide and, in this case, occurred with phocomelia of the upper limbs. Furthermore, these lower limb findings were clinically significant owing to the localized pain and physical dysfunction of the lower limb.

Case Report

A 54-year-old black female with a history of Müllerian anomaly of the uterus and phocomelia of the upper limbs (Fig. 1) due to in utero

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thalidomide exposure experienced recurrent, alternating right and left lateral foot and heel pain for a 6-year period (Table). The patient initially presented to her primary care physician in July 2010 because of a warm, swollen, and tender left ankle, which was localized to a 1.5-cm × 2-cm fluid-filled cyst on the left posteromedial heel. The cyst was aspirated with an 18-gauge needle during this visit. The patient also had a record of ganglion cyst removal in 1998. At the 2-month follow-up visit, a 1-cm cyst had recurred in the same location but without associated pain. However, the patient returned in August 2011 with pain and tenderness at the cyst site. The patient was prescribed oxycodone-acetaminophen 5 mg/325 mg orally for pain relief and referred to podiatry. The next week, the patient was found to have tenderness, swelling, and a palpable mobile mass at the left posterior medial heel. The patient underwent ganglion cyst removal under local anesthesia 1 month later.

In January 2014, the patient presented with severe pain ranging from dull to sharp and exacerbated by ambulation, prolonged standing, initial weightbearing, and particular footwear. During the episodes of foot pain, the patient experienced limited range of motion, numbness, and swelling of the foot. Initial radiographs showed dysmorphic changes to the ankle and foot, including foreshortening of the calcaneus and flattening of the talus (Fig. 2). Abnormalities were present in the articulation and narrowing of the talocalcaneal joint posteriorly, tibiotalar joint medially, and the fibulotalar joint. The articulating bony surfaces of these joints were also sclerotic, as were the talonavicular and cuneiform joints, consistent with degenerative joint changes of the ankle and midfoot. The source of pain was attributed to the hypermobility of the metatarsus with the lesser tarsus, coinciding with the diagnosis of metatarsalgia. Conservative treatment with naproxen 500 mg orally, orthotics, and methylprednisolone dose pack provided limited relief, and the patient continued to present regularly with foot pain. In May 2014, the patient presented with complaints of bilateral foot and ankle pain, with the right worse than the left, associated with flat fleet. The symptoms had been present for years but had recently progressed to constant pain with movement. The physical examination findings were specific for tenderness localized to the extensor digitorum longus tendon. Right tenosynovitis was diagnosed and treated conservatively with a stretching and icing regimen. During the next 2 years, the patient presented regularly with recurrence of alternating peroneal and calcaneal pain of the right and left lower extremities. Pain was reproducible with direct palpation of peroneal tendons and the intermediate dorsal cutaneous nerve as it crossed the rear foot and midfoot into the forefoot. Therefore, the patient underwent a trial of dexamethasone phosphate-triamcinolone acetonide steroid injections with 2% lidocaine and 5% bupivacaine into the left and right peroneal tendon sheaths and sinus tarsi in 2015. However, this treatment also failed to provide significant relief, similar to the results with the previous forms of conservative treatment.

Table
Signs and symptoms associated with lower leg/ankle anatomic variations

Symptoms and Signs
Patient-reported symptoms
Alternating lateral foot and heel pain
Pain with prolonged standing and ambulation
Numbness
Swelling
Stiffness
Clinically observed signs
Pes planus
Ganglion cyst

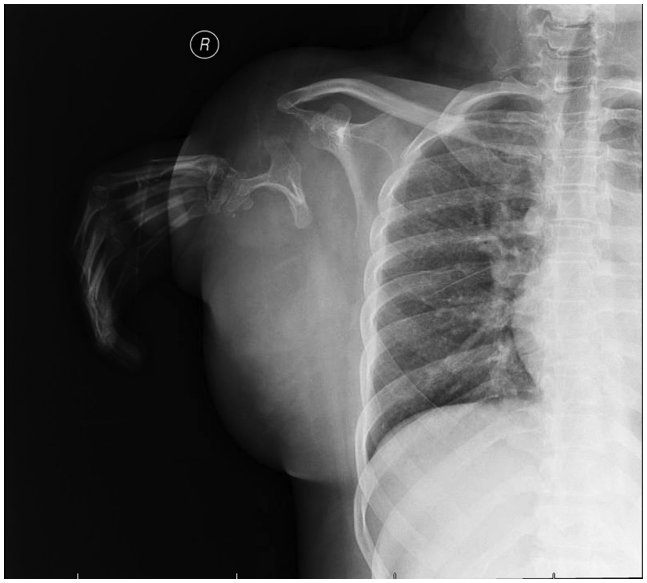

Fig. 1. Anteroposterior radiograph of the right shoulder revealing a markedly dysmorphic and foreshortened upper limb, classically associated with thalidomide exposure in utero.

In December 2016, magnetic resonance imaging (MRI) of the right ankle was obtained to further characterize the possible morphologic alterations associated with the patient's recalcitrant foot pain. A dysmorphic talus and calcaneus was again noted; however, several soft tissue anatomic abnormalities were also evident on the MRI study. The peroneus longus tendon was laterally dislocated, and the posterior tibialis and peroneus brevis tendons were absent (Figs. 3 and 4). Also, a variant tendon of the anterior compartment was present that inserted onto the calcaneus in the sinus tarsi (Fig. 3). Follow-up appointments in early 2017 addressed the high likelihood of the displaced and anomalous tendons contributing to the recurrent foot pain. Surgical release of these tendons was discussed as a treatment option; however, the patient chose to continue conservative treatment with meloxicam 15 mg orally and immobilization of the right foot with a controlled ankle motion boot.

Discussion

Variations in lower leg anatomy have been well-documented over the years with high-resolution imaging modalities such as MRI and ultrasonography facilitating identification of more of these abnormalities. Although often incidental findings, the identification of these abnormalities can, in certain cases, be clinically significant because they can be a source of chronic pain, gait abnormalities, and injury. In addition, some variants can be beneficial in surgical reconstruction. For example, Mick and Lynch (4) identified a peroneus quartus muscle during surgical repair of an avulsed superior peroneal retinaculum that was causing dislocation of the fibular tendons during dorsiflexion. This muscle was displaced forward and sutured in place to prevent recurrent fibular tendon dislocation.

The variant tendon identified in the present patient was located in the anterior compartment. The origin of the muscle was not visualized on MRI, but it coursed adjacent to the lateral aspect of the tibia and laterally to the extensor digitorum longus. The tendon inserted onto the calcaneus at the sinus tarsi (Fig. 2). The variant structure of the anterior compartment greatly resembled the muscle originally described by Lambert and Atsas (5) and Upadhyay and Amiras (6). Their cadaveric findings described anterior fibulocalcaneus, a structure that originates from the lower fibula and investing fascia of the peroneus tertius, tracking anteriorly to the lateral malleolus and inserting on the peroneal trochlea of the calcaneus. In concordance