Congenital Third Foot Deformity: A Case Report

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A congenital accessory limb is a rare condition that may be associated with predisposing factors such as drug or alcohol or toxin exposure, trauma, and deformities such as spina bifida. This article describes a case involving an accessory foot in an otherwise healthy 33-year-old male. It is likely that the etiology of this case was an early embryologic exposure to amniotic constriction bands that affected the development of the distal limb. Level of Clinical Evidence: 4 (The Journal of Foot & Ankle Surgery 47(6):583–588, 2008)

Key Words: accessory limb, amniotic band syndrome, ectopic limb, fetal abnormality, limb bud, limb duplication, mesoblastic fibrous strand, supernumerary limb

A congenital accessory limb is a rare condition, particularly in the absence of predisposing factors such as excessive drug, alcohol, or toxin exposure; obvious trauma; or associated deformities such as spina bifida. Vertebrate limbs first appear as a pair of bulges, or limb buds, originating from the thickened lateral plate mesoderm at the axial level of the neck-flank border and the flank-tail border in the developing embryo. The ectoderm surrounding this tissue thickens to form a specialized epidermal structure referred to as the apical ectodermal ridge. In 1925, Balinsky (1) reported that it was possible to "induce the formation of an additional limb" by transplanting other structures such as an ear vesicle to various locations on the flank. Similarly, ectopic limbs and limb structures can be induced in response to injury in experimental models that can undergo regeneration (eg, salamanders). In such cases, the induction and subsequent outgrowth of the ectopic limb is dependent on the presence of a viable nerve. The nerve dependency of both limb regeneration and ectopic limb formation has been investigated repeatedly, and several factors have been identified that may induce limb formation. One such factor is Fibroblast Growth Factor (FGF), which can rescue regeneration of denervated limbs, apparently through a mechanism involving the regulation of expression of the transcription factor, Dlx-3. The mechanism apparently involves the dedifferentiation of fibroblasts into a blastemalike structure (2). Lower extremity limb buds first appear around week 5 when the embryo is approximately 5 mm in length.

The initial blood supply to the lower extremity limb bud arises from an artery that eventually regresses. The common iliac artery initially gives off a branch, namely the external iliac. After this, the common iliac artery becomes the umbilical artery, a branch of which, known as the ischiadic artery, serves as the primary arterial trunk, or axis artery, of the developing lower limb. The ischiadic artery courses along the dorsal side of the thigh and leg. Below the knee, it runs between the tibia and the popliteus muscle. In the lower leg, it runs between the interosseous membrane and the tibialis posterior muscle, ending at the medial plantar aspect of the foot at the junction between the talus and calcaneus. It then gives off a perforating branch that traverses the sinus tarsi to initiate the blood supply of the dorsal arch. In the later stages of limb development, the ischiadic artery regresses and its function is taken over by the femoral artery, which is a continuation of the external iliac artery (Figure 1).

We hypothesized that the sequence of events that led to the development of the ectopic foot observed in the patient described in this article occurred during a period of time before which a new structure would easily form and after which a new structure would be unable to develop. Moreover, we anticipated that the ectopic foot would display a dedicated neurovascular supply, and share some portion of the connective tissues, namely muscle and/or tendon, with the adjacent limb.

Case Report

A 33-year-old male, of Hispanic descent and native to Mexico, presented with a cumbersome "third foot" on the medial aspect of his left leg, closely approximating the distal portion of the left Achilles tendon (Figures 2 and 3). With the exception of the chief complaint, he was healthy and athletic, and a good historian. The past medical history

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Financial Disclosure: None reported.
Conflict of Interest: None reported.

1067-2516/08/4706-0017$34.00/0
doi:10.1053/j.jfas.2008.08.008

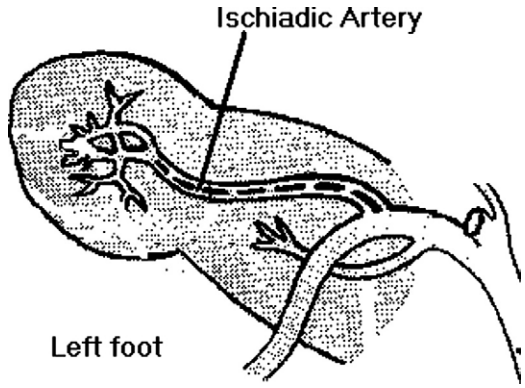

FIGURE 1 Limb bud week 5 (embryo approximately 5 mm). Modified from a diagram from the 35th British Edition of Gray's Anatomy, edited by Roger Warwick and Peter Williams, page 162 Figure 2.116, copyright ©Elsevier 1973.

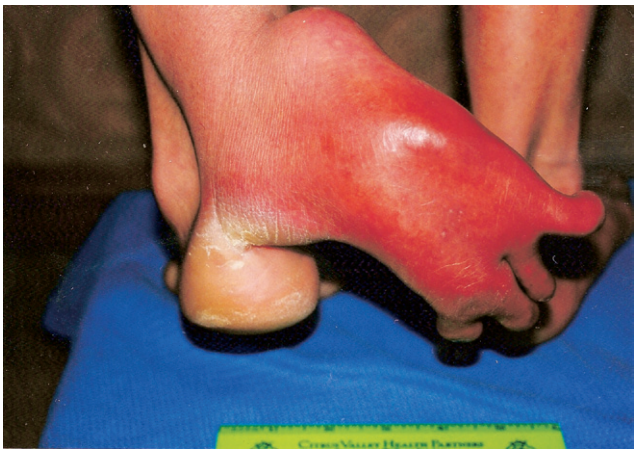

FIGURE 2 Ectopic foot posterior view.

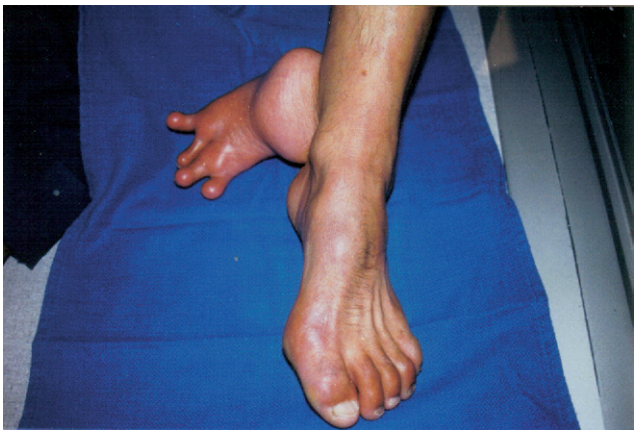

FIGURE 3 Ectopic foot anterior view.

was unremarkable, and he indicated that he was the product of a normal vaginal birth, and there was no history in the immediate family (including 7 siblings) of accessory appendages or other congenital abnormalities, or of mixed connective tissue disease, diabetes, or alcohol or drug abuse. The patient reported that his family initially sought medical intervention when he was 4 years old. He recalled being told after that examination that the physician told the family that the "false extremity" was "a bad omen," and that treatment would require amputation of the entire leg. For this reason, the patient never saw another physician in regard to the supernumerary foot.

Physical examination of the anatomic leg and foot (the normal left leg and foot) was remarkable in that it looked essentially normal. The size and shape of the anatomic left foot appeared to have developed symmetrically with that of the right foot. The accessory foot, attached to the anatomic left leg, was approximately 16 centimeters in length, and clinically appeared to be joined to the anatomic Achilles tendon via an 8-cm connection on the medial aspect of the leg near its distal attachment.

Based on clinical examination, including inspection of the ectopic limb for hair follicles and skin thickness, it was determined that the plantar surface of the accessory foot was facing in an anterior direction relative to normal anatomical position. Initial radiographs revealed the absence of an osseous articulation to the normal anatomical tibia or calcaneus outside of the accessory foot's anterior tibial and Achilles tendons (Figures 4 and 5). The accessory calcaneus was distal in its presentation and appeared to be fused to the accessory talus. There were 4 metatarsals, and phalanges were visible upon careful inspection of the radiographs. Manual muscle strength testing showed that the patient had weak voluntary control of the accessory foot and could accomplish a waving type motion. During ambulation, the patient had to swing his right leg outward in order to avoid hitting the accessory foot during swing through.

After discussion with the patient, it was clear that he wanted to eliminate the accessory foot if it could be done without having to amputate the entire left lower extremity. Based solely on the clinical and radiographic examinations, it was felt that the accessory foot could safely be removed and, after preoperative preparation of the patient, including a thorough informed consent process, surgery was undertaken. Under general anesthesia, with the patient supine on the operating table, and with a pneumatic tourniquet positioned on the left thigh, excision of the supernumerary foot was performed. The procedure was straightforward and relatively simple, and required approximately 34 minutes from start to finish. The approach was based on direct excision of the accessory foot using elliptical skin incisions that contained in the resultant wedge, the foot to be removed. Deep dissection revealed attachment to the anatomical Achilles tendon as well as a smaller attachment to the tibialis posterior tendon and muscle belly, leading into the dorsal (posterior) aspect of the accessory foot. A