Introduction

Diplopodia, or duplicated foot, is a rare congenital anomaly. It differs from polydactyly in that supernumerary metatarsal and tarsal bones are present as well as extra digits. Only a few cases of this anomaly have been reported in the literature to date. We present a newborn male without intrauterine teratogen exposure who was born with a duplicate foot of the left lower extremity and imperforate anus.

Case report

An infant male of nonconsanguineous parents was delivered by cesarean section following a full-term pregnancy complicated only by oligohydramnios. Routine prenatal ultrasound screening had shown no obvious abnormality. There was no history of maternal teratogen exposure, and there was no family history of diplopodia. Physical examination demonstrated a normal right lower extremity, and a fairly well developed duplicate foot emanating from the posterior-lateral aspect of the mid lower left leg (Fig. 1). The duplicate foot had four toes and appeared to have an Achilles tendon-like attachment to the leg. A strong dorsalis pedis pulse was palpated in the duplicate foot. No active motion could be elicited from the duplicate foot, but movement in the native left lower extremity was normal at all joints. Pulses and sensation were normal within the native left lower extremity. The baby also had a low imperforate anus that was surgically treated. No other physical abnormalities were detected.

Radiographic studies were done to evaluate the anatomy of both the normal leg and the duplicate foot (Fig. 2). The duplicate foot had four toes articulating with individual metatarsals. Two ossified tarsal bones, a calcaneus and talus, were also present. The muscular and vascular anatomy were further evaluated by MRI. Using a 1.5-T GE magnet, multiple T1- and T2-weighted acquisitions were obtained without and with gadolinium enhancement (Fig. 3). Additional MRA sequences were performed to identify the vascular anatomy prior to surgical amputation of the accessory foot (Fig. 4). The duplicate foot was attached from its calcaneus to the otherwise normal left lower extremity via an aberrant small accessory gastrocnemius muscle. A single artery, separate from the normal vessels of the calf, to the duplicate foot could be identified. While the origin of the artery supplying this foot was not delineated, it was identified in the upper thigh and clearly did not originate either at the superficial femoral or common femoral
arteries. It was therefore felt that the likely origin was from the iliac system.

The duplicate foot was surgically removed. At that time, an accessory gastrocnemius muscle was noted from the native calf with its Achilles-like tendon inserting into the accessory calcaneus. The neurovascular bundle was identified proceeding from the accessory gastrocnemius muscle to terminate at the medial first metatarsal base. Prior to closure, it was noted that the musculotendinous anatomy of the native left leg appeared otherwise normal, as did perfusion of the native foot. Dissection of the specimen revealed no muscular structures in the foot other than the aberrant gastrocnemius muscle described above.

Discussion

Limb formation begins at the end of the fourth week of embryonal development as small buds on the ventrolateral body wall; the lower limb buds appear shortly after the upper ones. The distal-most layer of ectodermal tissue, known as the apical ectodermal ridge (AER), induces formation of the limb by promoting growth and development, transforming the underlying mesoderm into what ultimately becomes the musculoskeletal system. The complex interplay between these various elements thus creates the potential for anomalies at multiple points in limb development with the most critical time period being days 24–36 after fertilization [4]. Early damage to the mesoderm or to the mechanisms of its instruction may result in absence or duplication of a structure and later malformations result in hypoplastic,remnant appendages. Supporting this, work by Sessions [5] in the frog Xenopus laevis showed that disruption of the developing native limb bud organization with implanted resin beads leads to multiple categories of limb deformities, including supernumerary limb structures.

Another hypothesis in limb development has suggested that an inciting event occurring before the seventh week of gestation gives rise to a spectrum of malformations, including conditions such as clubfoot, congenital fibular hypoplasia, tibial aplasia and diplopodia, all linked by an absence of the anterior tibial and dorsalis pedis arteries. It is thought this vascular aberration predisposes teratogenesis in the developing extremity [6]. While this theory may explain cases such as those reported by Karchinov [1], it is not supported by this current case. Our patient has a normal native anterior tibial and dorsalis pedis artery, as well as an accessory vessel from the level of the pelvis supplying the diplopodia.

This case is highly unusual in that complete duplication of the foot, save a single digit, occurred in an ectopic location; all reported cases of foot duplication in

Fig. 1 Photograph of the left leg in prone position in this infant with a duplicate foot

Fig. 2 (a) Frontal and (b) lateral radiograph of the left leg and duplicate foot shows that the duplicate foot has four toes, four metatarsals, and an ossification center for the calcaneus and talus
the English literature, to this point, have described diplopodia as part of the native foot. Furthermore, the vascular supply of the accessory foot was independent of the otherwise normal ipsilateral lower extremity. The added combination of diplopodia and imperforate anus has only been reported in one other case [3], in which there was prenatal thalidomide exposure. That patient also had ipsilateral proximal femoral focal deficiency, and absent upper limbs. Our case, however, had no maternal teratogen exposure. The early, prespecification event that must have occurred thus manifested as foot duplication without affecting the leg. Since the limbs develop in a proximal to distal fashion, the mechanism that allowed this to happen is unclear. Given the aberrant vascular supply to the duplicate foot originating proximally within the (probably) iliac system, one would have expected to see either duplication or malformation of the entire lower extremity rather than just a duplicate foot. Perhaps, as the aberrant vessel grew along the axis of the leg, it did not negatively influence induction of the surrounding mesoderm by the overlying AER. At some point in distal development, the vessel may then have split the limb bud, giving rise to the duplicate foot alone.

In summary, we present a very unusual case of a newborn boy with imperforate anus and diplopodia, with the accessory foot positioned posterolaterally at the left calf.

References