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Diplopodia with reversed foot

Normal gait after operation at 8 years of age

Diplopodia with the duplicated foot tucked up posteriorly, was reconstructed successfully in an 8-year-old boy. The knee joint, fibula and tibia were uninvolved; he had been walking on the dorsum of the foot wearing an ordinary shoe with the heel forward. The foot had ten toes, nine metatarsals and nine tarsal bones including one talus and one large calcaneus on which two Achilles tendons were inserted into two tuberosities. The lateral supernumerary foot was excised and the medial foot was dorsiflexed by almost 180° by massive release and elongation of tendons. He could walk on his bare reconstructed foot without a brace 10 months after the operation.

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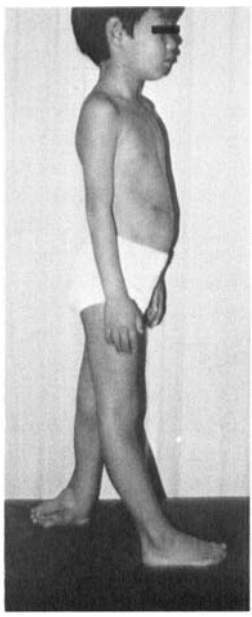
Diplopodia (duplicated foot), a rare anomaly mostly combined with hypoplasia or aplasia of the tibia, is treated by early resection of the supernumerary part of the foot and late stabilization of the knee joint and leg bones (Laurin et al. 1964, Karchinov 1973) or by Syme's or trans-knee amputation (Jones et al. 1978, Narang et al. 1982). We report a case of diplopodia which was successfully reconstructed at 8 years of age.

Case report

An 8-year-old boy was born with anal atresia and gross duplicated left foot. Amputation of the left foot was recommended at the age of 1 year, but was not accepted by the parents. He started walking on his deformed foot at the age of 2, wearing an ordinary shoe with the heel forward. He had been standing on the dorsum of the medial part of the foot and on the vertically oriented talar head which was covered by a gross callosity. He could run in an awkward manner but could not jump, and walking for a long distance caused pain from the callosity.

The left foot was duplicated and tucked up posteriorly. The hind foot was almost 90° in equinus and the forefoot was 180° plantarly flexed. Ten toes were aligned from the medial side (Figure 1); the first to fifth toes seemed to belong to the medial foot and the sixth to tenth toes to the lateral supernumerary foot. The feet faced each other at 45° and the lateral foot lay on the sagittal plane of the leg. All but the fifth, sixth and seventh toes could be moved actively. The foot could not be plantar- or dorsiflexed actively. There was no sensory deficit. The left kidney was missing. The karyotype was normal.

Radiographs showed one large calcaneus with two tuberosities. The medial foot contained one small talus, one navicular, three cuneiforms and one cuboid, while the lateral foot had only two tarsal bones between the calcaneus and metatarsals (Figure 2). All but the rudimentary fifth toe had metatarsal bones. Angiography revealed that the medial foot was supplied mainly by the medial and lateral plantar branches from the posterior tibial artery which ran posteriorly to the Achilles tendon and the calcaneal tuberosities. The lateral supernumerary foot was mainly supplied by one branch from the posterior tibial artery. Arthrography showed a well-preserved ankle joint extending posteriorly between the tibia and the displaced calcaneus.



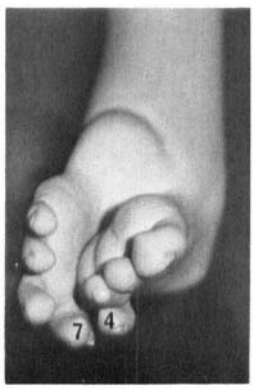


Figure 1. A. The patient standing on the dorsum of the left foot on a large callosity.
B. Posterior view of the left foot.

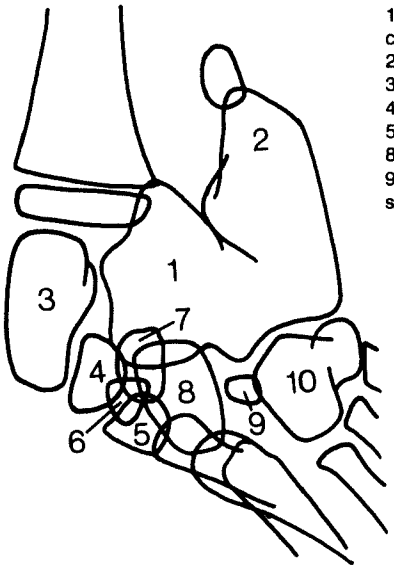


Figure 2. Lateral radiograph.
1, Medial tuberosity of the calcaneus.
2, Lateral tuberosity.
3, Vertically oriented talus.
4, Navicular.
5–7, Cuneiformis.
8, Cuboideum.
9, 10, Tarsal bones in the supernumerary lateral foot.

Operation

Part of the dorsal skin of the lateral foot was elevated as a large pedicle skin flap. Two short Achilles tendons, inserted into the two calcaneal tuberosities, were divided. When the foot was brought from extreme equinus to a neutral position, the posterior structures such as the posterior tibial tendon, short peroneal tendon and flexor hallucis longus tendon were divided transversely. The long peroneal tendon was left intact. The posterior capsulotomy was carried out widely to ease the reduction of the displaced talus. The lateral half of the calcaneus and two tarsal bones were excised. Although the medial foot could be reduced to a normal position, it was positioned at 30° of equinus and transfixed with Kirschner wires in case of undue tension to the posterior tibial artery. As it was impossible to elongate the short medial Achilles tendon, the proximal stump of the short peroneal tendon was transferred and fixed to the calcaneal tuberosity; the distal stump of the Achilles tendon was then sutured side-by-side to the short peroneal tendon. The posterior tibial tendon was sutured end-to-end to the distal stump of the flexor hallucis longus tendon. The flexor digitorum longus tendons were z-lengthened. The long peroneal tendon was translocated anteriorly over the lateral malleolus. The rudimentary 5th toe and the whole 8th, 9th and 10th toes with their respective metatarsal bones were excised. The 6th toe was excised, leaving the proximal half of the metatarsal bone. The 7th toe and distal half of the metatarsal bone were brought over to the 6th metatarsal bone, forming the new 5th toe.

The dorsal pedicle skin flap obtained from the excised foot covered the large skin defect over the heel. The foot maintained good circulation in the corrected position postoperatively although the skin flap over the heel became partially necrotic and was replaced by a skin graft after 2 months. Good bony consolidation was obtained.

A short leg brace was made with a rise of 4 cm and an external twist compensating the internal tibial torsion; a light night brace prevented recurrence of

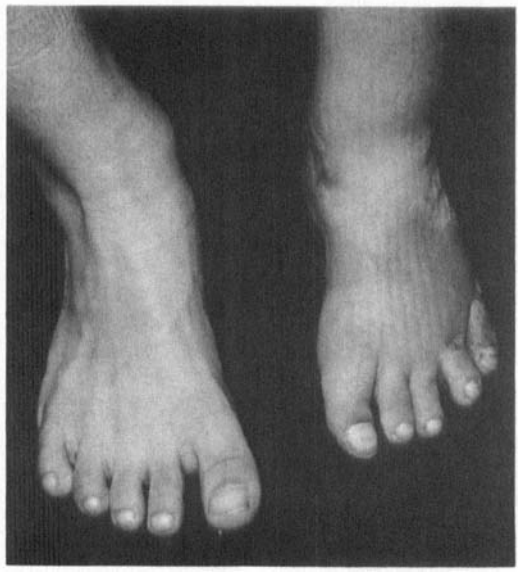


Figure 3. Four months after operation: the boy could stand on his bare left foot, and the callosity had disappeared.

the equinovarus deformity. When discharged 4 months after operation, the boy could stand on his bare left foot, and he had active plantar- and dorsiflexion of the foot and the first to fourth toes (Figure 3). The gross callosity on the dorsum of the foot had disappeared.

Discussion

Almost all reported cases of diplopodia have been associated with a defective tibia, except a case reported by Mysorekar & Lohokare (1970) who had a normal tibia and ankle joint although the foot was plantigrade and no mirror image was observed. Two Achilles tendons inserted into two calcanei were found by Narang et al. (1982). Jones et al. (1978) reported diplopodia in a male and his father, and a case associated with Thalidomide medication. Anal atresia was observed only in this Thalidomide case but the association of anal atresia with a defective tibia is known (Jones et al. 1978). The absence of one kidney has not been reported previously.

Diplopodia has been treated either by early resection of the supernumerary part followed by stabilization of the knee and leg bones, or by early amputation. Successful reconstruction at 8 years of age by rotating the plantarly flexed foot by almost 180° has not been reported previously.

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