



An Unusual Preaxial Polydactyly of the Foot; a Case Report

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ABSTRACT

Polydactyly is a commonly seen disorder. It can be seen as part of a syndrome or isolated deformity. Most commonly affects hand, foot or both but the presence of extra digit in only one foot is a very rare condition. We here present a very rare case of preaxial polydactyly of the foot with incomplete tarsal duplication, an accessory anterior tibialis tendon and without any flexor or extensor tendons in a 1-year-old male. The extra digit was removed successfully and the accessory anterior tibial tendon was repaired to the medial cuneiform. The purpose of this report was to discuss our results and to take attention to this rare presentation of polydactyly.

Keywords: Polydactyly; congenital foot deformity; child.

Ayakta Sıradışı bir Preaksiyel Polidaktili Olgusu; Vaka Sunumu

ÖZ

Polidaktili sık rastlanan bir deformitedir. Herhangi bir sendromun parçası olabileceği gibi izole bir deformite şeklinde de karşımıza çıkabilir. En çok el, ayak ya da her ikisini birden etkileyebilen polidaktilin sadece tek bir ayakta gözlenmesi oldukça nadir bir durumdur. Bu olguda, 1 yaşındaki bir erkek hastada gözlenen preaksiyel polidaktilin çok nadir bir formu ve tedavisi sunulmaya çalışılmıştır. Hastanın inkomplet tarsal duplikasyonu ve aksesuar anterior tibialis tendonu mevcut olup ekstra parmakta fleksör ya da ekstansör tendon bulunmamakta idi. Ekstra parmak başarılı bir şekilde çıkarıldı ve aksesuar anterior tibial tendon medial küneiforma taşındı. Bu çalışmanın amacı sonuçlarımızı tartışarak nadir gözlenen bu polidaktili formuna dikkati çekmektir.

Anahtar Kelimeler: Polidaktili; doğuştan ayak şekil bozukluğu; çocuk.

INTRODUCTION

Polydactyly is a common disorder of newborns with an incidence of about 0.2 % in live births and it can be seen as part of a syndrome or isolated (1). The condition can affect hand, foot or both but the presence of extra digit in only one foot is a very rare condition. Classification is traditionally made as preaxial, postaxial and central (2). Preaxial polydactyly of the foot is a rare entity accounting for about 15% of the foot polydactyly cases (3). Morphologic classification can be done either by Venn-Watson (4) or Watanabe (5) classification systems. We present a case of preaxial polydactyly of the foot with incomplete tarsal duplication.

CASE REPORT

A 1-year-old male was presented to our Orthopedics outpatient clinic with complaints of an extra first digit of left foot. The child was able to ambulate without any difficulty but he could not wear shoes comfortably. There was not any history of polydactyly in the family and consanguinity between the parents. The child was healthy and no history of any developmental delay was noted.

Physical examination of the child revealed a preaxial polydactyly of the left foot (Figure 1). The other digits of the foot were normal by inspection. After a detailed examination, it was noted that there was not active dorsiflexion in the normal hallux but the extraneous digit was having. Roentgenographic evaluation of the foot was done (Figure 2) and revealed a tarsal type preaxial polydactyly according to Watanabe classification system.

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Figure 1. Preoperative photograph of the left foot showing preaxial extranumerary hallux. The normal hallux was hypoplastic with slight plantar flexion deformity.



Figure 2. Preoperative radiographic examination of the left foot showing an incomplete tarsal type preaxial polydactyly.

The child was operated under general anesthesia. A medial fish-mouth incision was used for resection. There was an accessory anterior tibialis tendon (ATT) inserting at the base of the extranumerary digit (Figure 3) but no flexor tendon was noted. The detailed exposure of the extensor mechanism of the hallux revealed no extensor hallucis longus (EHL) tendon.



Figure 3. Intraoperative photograph showing an accessory anterior tibial tendon inserting at the base of extranumerary digit.

The accessory ATT was resected from the insertion point and it was sutured to the medial cuneiform (Figure 4). Then the incision was remodeled and closed by absorbable No. 5/0 suture (Figure 5).



Figure 4. The accessory anterior tibial tendon was sutured to the medial cuneiform.



Figure 5. Postoperative appearance of the sutures and reconstruction.

A lower leg cast was applied to the child to help the tendon healing. After 6 weeks from the surgery, the cast was removed and the child was allowed to walk. Antero-posterior and lateral X-rays of the foot were taken at the 6th week of surgery there was not any early complication on this views (Figure 6).

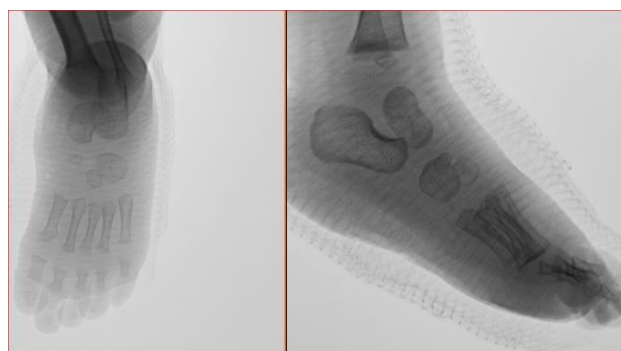


Figure 6. Postoperative radiographic examination of the left foot at the 6th week after surgery.

DISCUSSION

Isolated and non-hereditary preaxial polydactyly of the foot affecting only one extremity is a very rare condition as in our case. Reported studies show that polydactyly is hereditary in most of the cases and affects both hands and feet often with contralateral distribution (6). The most prevalent type of polydactyly is postaxial and there are

limited studies describing the preaxial polydactyly (3,7,8). In a study by Belthur et al. comparing the classification systems of polydactyly of the foot, the Watanabe classification was found to be more useful than Venn-Watson system (9). A tarsal type (true prehallux) preaxial polydactyly was seen in our case according to Watanabe classification system which is seen with an incidence of 3% in all the preaxial polydactyly cases (5). In our case the extraneous digit was developed incompletely but had an accessory ATT with active dorsiflexion movement in the digit. The condition is called as true prehallux and first described by Cobey et al. in 1966 (10). After this time, there are only a few reported cases of this condition in English literature (9). In addition to presence of an accessory ATT, the child in our case was lacking EHL muscle and the normal hallux had no active dorsiflexion. Surgical repair of the EHL tendon to avoid a functional deficit of the foot have been advised (11). The parents of the child were informed about the deformity of the hallux, repair of the EHL tendon and the problems that may occur if the repair is not performed; but they stated that they accept these risks and did not want to perform an extended repair surgery. The surgical reconstruction of the foot in polydactyly patients is recommended to be done at walking age for good shoe wear fit (4). The most commonly encountered complication and the cause of poor outcomes after preaxial polydactyly is recurrent hallux varus (9,12). To overcome this complication, the excision of the most medial digit is recommended. Because the extra digit in our case was tarsal type and did not affect the normal hallux and 1st metatarsal, no hallux varus was encountered before and after the surgery.

CONCLUSION

Although the preaxial polydactyly of the foot is a very rare condition, none of the reported cases have discussed about EHL lacking and to our knowledge this is the first study discussing this entity. None of the polydactyly cases are identical and the surgeon must be alert to the possibility of encountering different situations while dealing with polydactyly cases.

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REFERENCES

1. Phelps DA, Grogan DP. Polydactyly of the foot. *J Pediatr Orthop.* 1985; 5(4): 446–51.
2. Chiang H, Huang SC. Polydactyly of the foot: manifestations and treatment. *J Formos Med Assoc.* 1997; 96(3): 194–8.
3. Haber LL, Adams HB, Thompson GH, Duncan LS, Didomenico LA, McCluskey WP. Unique case of polydactyly and a new classification system. *J Pediatr Orthop.* 2007; 27(3): 326–8.
4. Venn-Watson EA. Problems in polydactyly of the foot. *Orthop Clin North Am.* 1976; 7(4): 909–27.
5. Watanabe H, Fujita S, Oka H. Polydactyly of the foot: an analysis of 265 cases and a morphological classification. *Plast Reconstr Surg.* 1992; 89(5): 856–77.
6. Gawlikowska-Stroka A. Polydactyly and syndactyly as the most common congenital disorders of the limbs. *Ann Acad Med Stetin.* 2008; 54(3): 130–3.
7. Granite G, Herzenberg JE, Wade R. Rare case of tibial hemimelia, preaxial polydactyly, and club foot. *World J Clin Cases.* 2016; 4(12): 401–8.
8. Moore JL, Joseph A. Complete First Ray Polydactyly: A Case Report. *J Foot Ankle Surg.* 2018; 57(5): 1027–9.
9. Belthur MV, Linton JL, Barnes DA. The spectrum of preaxial polydactyly of the foot. *J Pediatr Orthop.* 2011; 31(4): 435–47.
10. Cobey MC, Cobey JC. A true prehallux. The first to be described in the literature. *J Bone Joint Surg Am.* 1966; 48(5): 953–4.
11. Scaduto AA, Cracchiolo A III. Lacerations and ruptures of the flexor or extensor hallucis longus tendons. *Foot Ankle Clin.* 2000; 5(3): 725–36.
12. Turra S, Gigante C, Bisinella G. Polydactyly of the foot. *J Pediatr Orthop B.* 2007; 16(3): 216–20.