

Successful intervention of postaxial polysyndactyly beyond infancy. A case report

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Case Report

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Background: Polysyndactyly is a congenital anomaly involving both supernumerary digits and syndactyly. We report a case of a middle childhood female presenting with postaxial type B polysyndactyly and syndactyly of the fourth and fifth toes on the right foot. Radiographs confirmed a supernumerary digit fused distally to the fifth toe. The patient experienced discomfort when wearing shoes. Surgical correction included syndactyly release and excision of the non-functional fifth toe using local flaps, avoiding the need for skin grafts. Postoperative recovery was uneventful, with full functional and cosmetic restoration. This case highlights the importance of individualized surgical planning based on anatomical dominance, neurovascular preservation, and flap technique to achieve favorable outcomes even when surgery is delayed beyond infancy.

Keywords: Postaxial Polysyndactyly, Supernumerary Toe, Syndactyly, Polydactyly.

Polysyndactyly is a congenital malformation that occurs in the hands, feet, or both at the same time and consists of the existence of multiple supernumerary fingers¹.

Polysyndactyly is an autosomal dominant genetic limb malformations characterized by incomplete penetrance and phenotypic variability, and can occur independently or as part of a syndrome. Several gene mutations, including *HOXD13*, *GLI3*, *GJA1*, *FBN1*, *Lmbr1* and *SHH*, were verified as direct causes of polysyndactyly².

Among limb malformations, polydactyly represents the most common congenital deformity with an estimated incidence of 1/700–1000 live births³.

Polydactyly refers to the partial or total presence of supernumerary digits. The duplication can be preaxial or internal (duplication of the first digit), postaxial or external (affecting the fifth digit, accounting for 80% of foot polydactylies), or central (affecting the second, third, and fourth digits, and being occasional)⁴.

Postaxial polydactyly is further classified as type A and type B. In type A, the severe type, the supernumerary digit is well developed and articulates with either the fifth or a supernumerary metacarpal/metatarsal. In type B, the milder type, the supernumerary digit is rudimentary and feeble, and may even appear as a small protuberance, tag, or pedunculated nubbin. The relative proportion of types A and B varies among populations⁵.

Syndactyly is a congenital digital malformation characterized by webbing of the fingers and toes. It is derived from the Greek word “syn”, meaning together, and “dactylos”, meaning digits⁶.

It has a prevalence of 3–10 in every 10,000 births. Its occurrence in males is twice that in females, and mothers aged 40 years or older are more likely to produce children with inborn limb deformities compared to mothers who are 30 years of age or younger⁶.

They may be an isolated entity (possibly related to a genetic etiology), as component than three hundred hereditary syndromic malformations or as component of an amniotic band syndrome. Soft tissues can be only affected or be associated with a bone or cartilage abnormality⁷.

For newborns with simple syndactyly, the best age to receive surgery ranges from 6 to 18 months old, whereas in case of complex syndactyly, surgery should be performed prior to 6 months of age⁶.

Surgical treatment involves a risk of recurrence (28%) and potential complications (11,7%)⁷.

A small percentage of patients experience postoperative keloid formation around the incision area, which may result in pain and pruritus and even affect shoe-wearing and foot movement⁸.

When the condition involves syndactyly of the fourth and fifth toes, it is referred to as polysyndactyly, which is characterized by a dominant



Figure 1. Postaxial Polysyndactyly type B

fifth toe and a hypoplastic sixth toe⁹. The goal of treatment for postaxial polydactyly is to achieve a well-supported, contoured foot that is compatible with normal footwear. Ideally, treatment should be performed before the first year of age to avoid difficulties with footwear before the onset of walking⁴.



Figure 2. Foot radiograph: AP and oblique view



Figure 3 (left): Plantar incision design. **Figure 4** (right): Dorsal incision design

Simple excision of the extra toes does not yield satisfactory results due to the diverse and complicated shapes of polysyndactyly of the foot. The choice of which toe to excise has a significant effect on the postoperative outcome and cosmesis, and remains a controversial issue⁹.

The commonly described treatment for postaxial type polysyndactyly involves resection of the fifth toe to preserve the neurovascular bundles of the fourth and sixth toes⁹.



Figure 5. Distal phalanx resection



Figure 6. (left) Immediate post surgery. **Figure 7.** (right) 3 days post surgery

Case report

A female patient in middle childhood with no relevant medical history presented with syndactyly of the 4th and 5th toes and postaxial polysyndactyly of the right foot (Figure 1), causing mild discomfort while wearing shoes due to spatial compromise.

Anteroposterior and oblique radiographs of the right foot (Figure 2) revealed a supernumerary toe with fusion distal to the 5th toe.

Surgical correction was planned. After a fifth radial nerve block with bupivacaine and standard surgical preparation, supramalleolar ischemia was induced with a hemostatic cuff. A figure-of-eight (lemniscate) incision was made on the plantar aspect between the fifth and supernumerary toes (Figure 3), along with a vertical incision between the fourth and fifth toes (Figure 4).

Dissection proceeded through the dorsal incision to separate the syndactylized fourth and fifth toes. Further dissection around the fifth and supernumerary toes exposed the proximal interphalangeal joint capsule. Capsulotomy and release of the collateral ligaments allowed for osteotomy of the distal phalanx (Figure 5).

Closure was achieved using layered suturing of local flaps (Figure 6). The postoperative outcome was favorable.

The patient had no complications, and follow-up visits at 3 (Figure 7), 7, and 14 days showed satisfactory wound healing. Sutures were removed at two weeks, and the patient was discharged without complications or residual discomfort.

Discussion

This case illustrates the surgical decision-making process in a middle childhood patient with postaxial polysyndactyly and associated syndactyly.

Polysyndactyly is one of the most common congenital limb deformities and is associated with a complex genetic background involving multiple genes.

Among these, mutations in the HOXD13 gene have been widely implicated².

In a review of 147 cases by Miguellas, 52 patients had a relevant family history¹. In our case, the patient reported a family history of polydactyly in an aunt, supporting a potential hereditary component. Although genetic testing was not performed in this case, it is worth considering in similar presentations to better understand underlying syndromic or familial patterns.

Family-based genetic studies have identified various loci associated with postaxial polydactyly, such as the GLI3 gene on chromosome 7p, PAPA2 on chromosome 13q, and PAPA3 on chromosome 19p¹⁰. These findings underline the complexity of the genetic mechanisms involved.

Surgical correction remains the mainstay of treatment for polysyndactyly and syndactyly, especially when symptomatic or cosmetically concerning. In our case, the indication was mild pain and footwear intolerance due to spatial constraint from the supernumerary toe. Various flap designs have been proposed to manage syndactyly without requiring full-thickness skin grafts, aiming to reduce donor site morbidity and improve aesthetic outcomes. Some studies also report the potential for spontaneous epithelialization of skin defects⁷. In this case, the wound was closed with local flaps, without the need for grafting, and there were no complications such as flap necrosis or keloid formation. There are currently no evidence-based clinical guidelines to standardize the timing or technique of surgical correction in such cases. Although the literature presents a range of surgical approaches and timings, no clear consensus has been established³. This underscores the need for individualized surgical planning.

A key surgical decision in postaxial polysyndactyly is determining which toe to excise. Resection of the fifth toe is often preferred to preserve the lateral neurovascular structures of the sixth toe¹¹. In our case, we followed this approach due to the neurovascular advantage and better cosmetic potential. The sixth toe had better alignment and structure, while the fifth toe was supernumerary and non-functional.

Moreover, preserving the sixth toe avoids scarring on its lateral surface and facilitates reconstruction of the fourth interdigital space¹¹.

The SAM classification provides guidance for decision-making in cases with axis deviation or hypoplasia. In types A2 and A3, where the sixth toe is short and deviated, preserving the fifth toe has shown better functional and cosmetic results. Our patient did not exhibit such deviation, supporting the surgical choice made¹².

Although early intervention (within the first years of life) is often recommended, our case

demonstrates that despite the late intervention, a favorable aesthetic and functional outcome was achieved using local flaps without grafts, which supports the feasibility of such approaches in selected cases.

The patient experienced relief from discomfort, improved tolerance to footwear, and expressed satisfaction with the cosmetic appearance, which positively impacted her self-confidence.

This case reinforces the importance of individualized surgical planning in polysyndactyly, particularly in patients presenting beyond infancy. Further comparative studies are needed to determine the long-term benefits of different flap techniques and timing of intervention.

Conclusion

Polysyndactyly of the foot is a common congenital malformation, often with a hereditary component. Although preventive strategies are not available, early surgical intervention can prevent deformities and improve outcomes.

In this case, surgery was performed beyond the ideal age, yet successful results were achieved by combining syndactyly separation with excision of the supernumerary toe. Precise identification of the less functional toe, typically the fifth, is essential for optimal results.

This case emphasizes the importance of individualized surgical planning, considering patient age, anatomical features, genetic background, and surgeon expertise. Further case series and long-term studies are needed to inform standardized treatment protocols and improve care.

Conflicts of interests

None declared by the authors.

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