

Operative Management of Y-Shaped Metatarsal with Biphalaengeal Sixth Toe

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ABSTRACT

BACKGROUND: In the literature, there is no consensus regarding the surgical management of postaxial polydactyly, and few cases of polymetatarsia with polydactyly have been reported. Treatment of the complete deformity will prevent further foot and gait disorders.

OBJECTIVE: To identify literature relevant to the operative management of Y-shaped metatarsal with biphalaengeal sixth toe and related skin and wound care to improve surgical treatment protocols from a clinical experience perspective.

DATA SOURCES: The authors searched several electronic databases in December 2022 for articles related to postaxial polysyndactyly in the feet and polymetatarsia. Databases searched included PubMed, SciELO, ScienceDirect, Cochrane Database of Systematic Reviews, and Google Scholar gray literature.

STUDY SELECTION: Two independent researchers conducted the searches and read the article titles and abstracts. Studies were included if they were narrative reviews, case studies, or observational studies; written in English or Spanish; and published between 2012 and 2022. Nonhuman studies were excluded. Studies that met the inclusion criteria were fully evaluated. Disagreements between reviewers were resolved by consensus, and when there was no consensus, a senior researcher was consulted.

DATA EXTRACTION: The following data were extracted from the included studies using a standardized form: author and year of publication, study type, number of participants, sex, polydactyly location, polymetatarsia, type of polydactyly, participants' history of hereditary associated diseases or malformations, treatment, removal criteria, and timing of surgery.

DATA SYNTHESIS: Authors evaluated 11 studies of postaxial polydactyly that included a total of 153 participants (64 men, 89 women). They also document their clinical experience with a surgical technique used in cases of bilateral postaxial polydactyly of the foot with a Y-shaped metatarsal with biphalaengeal sixth toe.

CONCLUSIONS: Surgical correction with lateral removal of the sixth toe is a resolutive treatment to improve the functionality of the foot, its aesthetic appearance, and the patient's quality of life. Case-specific treatment should be applied and tailored to meet the individual needs. The biomechanics of gait and shoe problems in these patients improve with surgical treatment, without presenting secondary aesthetic problems in skin care.

KEYWORDS: foot, malformation, metatarsal, polydactyly, polymetatarsia, surgery

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INTRODUCTION

Polydactyly is the most common congenital malformation of the forefoot; it can be partial (phalange or metatarsal) or complete with a developed accessory radius and toe duplication. Postaxial polydactyly is defined as a complete or partial supernumerary toe on the lateral aspect of the foot; this constitutes 80% of duplications. The condition affects both sexes equally, but it is more prevalent in certain ethnic groups. Rates are highest in African and American populations. That said, in the scientific literature, Nigeria, Japan, and the Netherlands are the regions that have most commonly reported this condition.¹ In Japan, polydactyly surgery is a common choice because of the cultural preference for going barefoot. Some authors report biphalaengeal fifth toes more frequently in those of Danish, French, or Swedish descent than in African Americans.¹ In Spain, Gallart et al² studied 2,494 feet and found that 53.7% of the fifth toes were triphalaengeal and 46.3% were biphalaengeal toes. They also reported that triphalaengeal fifth toes seem to present a higher proportion of pathology.²

That said, most extant studies do not stratify outcomes according to the level of duplication (metatarsal vs phalaengeal). The majority of reports excise the nondominant ray, regardless of the level of duplication. Among the few studies focusing on biphalaengeal rather than metatarsal duplication, Chocron et al³ reported a total of 127 cases: 100 cases of biphalaengeal and 27 cases of metatarsal duplication. Interestingly, Yuan et al⁴ reported a case of triphalaengeal fifth toe in a 31-year-old Asian woman. However, it is more common to find biphalaengeal fifth toes described in the literature.

There are different manifestations of polydactyly,⁴ and it may be associated with other diseases or syndromes (Figure 1), including VACTERL (vertebral defects, anal atresia, cardiac defects, tracheo-esophageal fistula, renal anomalies, and limb abnormalities) association, Fanconi anemia,

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Figure 1. POSTAXIAL POLYSYNDACTYLY ASSOCIATED WITH TRISOMIC DISEASE



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and trisomy diseases (with associated abnormalities of the *GLI3* and *SHH* genes).^{3,5} Several classifications of polydactyly have been described in the literature, some based on morphology and anatomy⁶ and others, such as the classification by Lee et al,⁷ founded on morphologic and radiographic appearance. Other authors report that polydactyly in the lower limbs can be tibial, peroneal, or central.⁶

In most cases, this malformation has little impact on the patient's ability to walk. Accordingly, there is no clear consensus on a treatment guideline for the malformation, including whether treatment is necessary. Some patients are reluctant to undergo surgery and are treated with plantar supports and shoe modifications (Figure 2). Critically, there is a lack of consensus on an ideal approach to surgical management.^{8,9} This controversy is greatest when deciding on the appropriate age at which the patient should undergo surgery.¹ Despite this controversy, several authors have proposed different classifications of polydactyly malformation to facilitate management and surgical approach.¹⁰⁻¹² Pain, biomechanical problems of mobility and function, difficulty with footwear, sensitivity when walking, and aesthetic concerns are the factors that can influence the decision to undergo surgery (Figure 3).^{9,13}

Excision of the supernumerary toe must meet certain requirements: it must be the toe with the greatest aberration and with the least functionality and anatomical development, and deficits must relate to the stability and biomechanics of the forefoot.¹⁴ Most literature reports satisfactory results for patients treated surgically. However, patients should be followed up for approximately 6 to 12 months after surgery and be evaluated for residual or future deformities that may occur. In the case of fibular polydactyly, there may be some subsequent complication of angular deformity, but the resulting scar is a positive outcome variable of treatment. Few cases of

polymetatarsia with polydactyly have been reported in the literature.

Given all of these factors, the authors conducted a review of the existing scientific literature on postaxial polysyndactyly and polymetatarsia, with the goal of improving surgical treatment protocols from a clinical experience perspective. The authors compiled the resulting data into an overview to enable deeper perusal for clinicians and open this topic for future investigation and academic discussion.

METHODS

This article was not intended to be a systematic review or PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses)-guided search. For this narrative review, the authors analyzed several electronic databases including PubMed, SciELO, ScienceDirect, Cochrane Database of Systematic Reviews, and Google Scholar gray literature. All the included studies were published between 2012 and 2022. The searches were conducted in December 2022, after building the strategies used in the different databases.

Keyword searches were assigned to Medical Subject Headings (MeSH). The following search strings were used: (polydactyly AND foot); (therapy and polydactyly and foot) AND surgery); (postaxial polydactyly AND foot). Narrative reviews and case studies, including observational studies, that addressed polydactyly and foot surgery were included. Articles were required to be published in English or Spanish. The authors excluded studies that were not conducted in humans.

Two independent reviewers carried out the selection procedure. Each reviewer read the title and abstract of

Figure 2. PATIENT WHO REFUSED TREATMENT OF FOOT SURGERY



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Figure 3. DERMAL LESIONS SECONDARY TO POLYDACTYLY

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each paper and assessed whether each study met the inclusion criteria. Selected articles were screened again by the other reviewer. Full texts were assessed for their suitability according to precise compliance with the defined inclusion criteria. The following data were extracted from each study using a standardized form created by the researchers: author and year of publication, study type, number of participants, sex, polydactyly location, polymetatarsia, type of polydactyly, participants' history of hereditary associated diseases or malformations, treatment, removal criteria, and timing of surgery.

During data extraction and to simplify study selection given the high number of references analyzed, an Excel (Microsoft Corp) document was designed as a data collection form through which inclusion was assessed by coding criteria. Occasionally, the mediation of a third reviewer was required to decide the final inclusion or exclusion of some of the publications. Figure 4 shows the flowchart used to represent the different phases of the selection procedure of the studies finally included in the review.

RESULTS

The authors identified 274 studies; ultimately, 11 studies were included. The total number of participants in all the studies was 153 (64 men, 89 women). The studies were conducted on children, adolescents, and adults. The Table presents the main characteristics of the studies and the most significant data extracted in this review.

The main findings of this review are that the clinical and surgical conditions of polydactyly with and without polymetatarsia require different treatment, and surgical treatment must be personalized. There exists a paucity of guidelines on the surgical management of polymetatarsia.²⁵ The presentation of postaxial polydactyly is very varied and may require different treatment modalities.

CASE REPORT

In this case report, the authors document a surgical technique used in a patient with bilateral postaxial polydactyly of the foot with a Y-shaped metatarsal with biphalangal sixth toe (Figure 5). The authors describe the method of surgical correction and the need for preoperative planning to achieve a good functional and cosmetic result and improve the patient's quality of life.

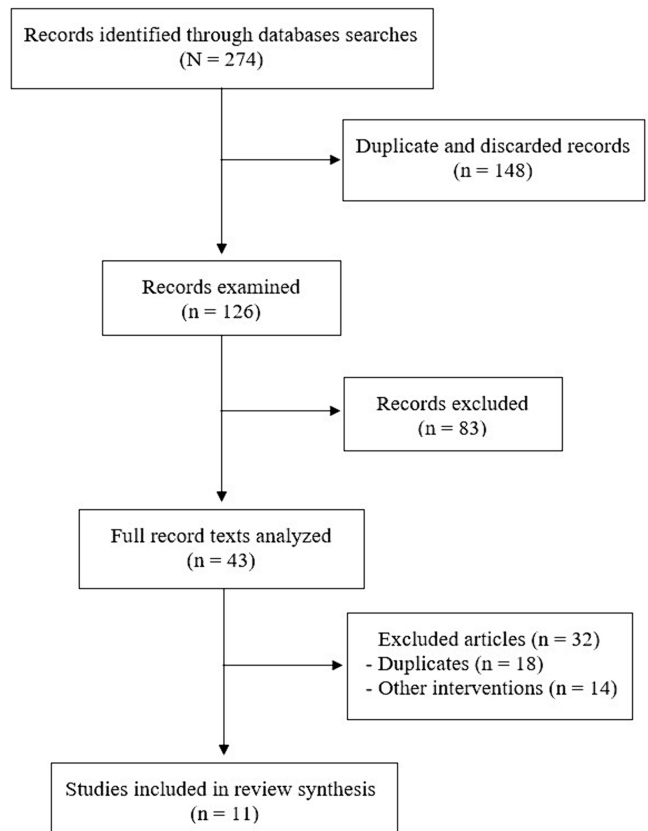
Ethical Considerations

The patient was informed that data concerning the case would be submitted for publication, and he provided consent. The ethical standards for human research stipulated in the Declaration of Helsinki (World Medical Association), in the Council of Europe Convention on Human Rights and Biomedicine, in the UNESCO Universal Declaration on the Human Genome and Human Rights, and in similar institutional declarations were observed at all times. The case has the authorization of the clinical podiatry unit of the university. Participation was voluntary, and no incentives were offered for participation.

Physical Examination and Clinical Features

In this clinical case, the patient was born with a bilateral sixth toe without an association with any other genetic

Figure 4. STUDY SELECTION FLOWCHART



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Table. SUMMARY OF INCLUDED STUDIES

Authors	Participants; Sex; Side	Polymetatarsia	Type of Polydactyly	Hereditary	Associated Diseases or Malformations	Treatment	Removal Criteria	Timing of Surgery
Bingol et al, ¹⁴ 2021	1; male; left	Yes	Postaxial	No	No	"On-top plasty" method	Direct excision of an additional finger would not be sufficient	-
Burger et al, ¹⁵ 2018	76; male: 30, female: 46; right: 15, left: 6, bilateral: 55	Yes	Preaxial	Yes: 41; no: 32; unknown: 3	Lower limb: 55; upper limb: 59; craniofacial: 32; genetic mutations: 27	-	-	-
Cheng et al, ¹⁶ 2021	1; female; bilateral	Incomplete polymetatarsia with a T-shaped metatarsal head in the left foot and a Y-shaped metatarsal head in the right foot	Postaxial	No	No	Removal of the medial toe	Evaluation is individualized - considering circulation, site of origin, presence of hypoplasia, fusion status, axial alignment, and general appearance	-
Escudero et al, ¹⁷ 2017	1; female; bilateral	No	Postaxial	No	-	Medial fifth-toe excision, plantar plate repair, and medial collateral ligament reconstruction were performed due to malformation and widening of the distal surface of the plantar plate	Preservation of lateral supernumerary toe	Early childhood
Han et al, ¹⁸ 2014	1; male; right	Yes	Postaxial	No	No	Bone transfer of the extra metatarsal bone and subsequent fixation with an internal plate	Normalization of the metatarsophalangeal joint and the level of this joint along with alignment and cosmesis.	-
Hiraoka et al, ¹⁹ 2020	1; female; right	No	Central	No	A separated toenail	Resection of the lateral side of the phalanx and fixation of the medial side to a medial distal phalanx with a Kirschner-wire	Preserve the medial side of the second distal phalanx because it was larger and in good alignment with the middle phalanx	1 y of age
Kubat and Antičević, ²⁰ 2018	30 feet, 24 patients; male: 8, female: 16; bilateral: 6, unilateral: 18	No	Postaxial: 16; preaxial: 8	No	Oral-facial-digital syndrome and hand syndactyly: 1; Ellis-van Creveld syndrome: 1; hallux varus deformity: 2; left iliac bone hypoplasia: 1	Surgery involved saving the most dominant ray and digit	The medial ray was preserved and the lateral one excised in most cases of postaxial polydactyly; patients with preaxial polydactyly usually had the medial toe removed along with the corresponding metatarsal bone, if present	Timing of surgery is not crucial for final results
Oragui et al, ²¹ 2012	1; female; right	Yes	Postaxial	No	No	Medial rudimentary digit was excised at the fused base of the phalanx	Surgical treatment is individualized	Delay skeletal correction until patient is 1 y

(continues)

Table. SUMMARY OF INCLUDED STUDIES, CONTINUED

Authors	Participants; Sex; Side	Polymetatarsia	Type of Polydactyly	Hereditary	Associated Diseases or Malformations	Treatment	Removal Criteria	Timing of Surgery
Park et al, ²² 2013	36 feet, 27 patients; male: 16, female: 11; bilateral: 9, unilateral: 18	Yes: 18	Postaxial	No	No	To excise the medial toe, a dorsal rectangular flap, and a full-thickness inguinal skin graft. To excise the lateral toe, a racket-shape incision	Morphology (size, appearance) and radiologic configurations (hypoplasia and additional toe deviation from foot longitudinal axis). Excision can achieve a foot with a natural contour and a deep natural web space	
Piette et al, ²³ 2017	1; female; left	Yes	Central	No	No	Removal of the second and third rays and reconstruction of the intermetatarsal ligament of the first and fourth metatarsals	Removed the most misaligned toes	Until 1 y of age when foot anatomy has developed
Yamada et al, ²⁴ 2012	19; male: 8, female: 11; unilateral	No	Postaxial polysyndactyly	No	No	Designed a rectangular flap on the tip of the sixth toe and a triangular flap on the dorsum of the sixth toe; removal of the sixth toe	Hypoplastic toe (in all cases, the fifth toe was dominant and the sixth toe was hypoplastic)	

disease. He reported no family history of this malformation. His main complaint was bilateral pain in both feet and difficulty walking. He also reported an abnormal cosmetic appearance and difficulty wearing shoes. The supernumerary toes made it impossible for the patient to wear standard shoes. These reasons supported a surgical resolution in adulthood, as the patient's parents had been concerned and fearful about performing surgery during childhood. The patient had not received conservative treatment with foot orthoses. Over the patient's lifetime, any calluses or foot wounds (erosions, lacerations, hematomas, nail lacerations, and friction blisters) had been treated by nurses.

Physical examination revealed fully developed extra toes on both feet and dermal lesions on the lateral border of the foot related to cramped footwear. The patient was in the habit of regularly visiting his podiatrist for podiatric treatment of these skin lesions. However, these foot problems increased over time, and finally, the patient was referred to the authors' medical center for surgery.

Providers ordered standard anterior-posterior and lateral radiographs of the patient's feet to provide a better picture of the extent of metatarsal involvement and deformity. Radiology showed a Y-shaped bifurcation of the fifth metatarsal head with complete duplication of proximal and distal phalanges of the fifth and sixth toes (Figures 6 and 7). Further, the metatarsals presented as incomplete forms of polymetatarsia. The fifth

metatarsophalangeal joint was large, with a more pronounced lateral deviation in the right foot.

Surgical Technique

Providers placed the patient in the supine position and provided anesthesia by means of a truncal block to both feet with 0.75% bupivacaine. A tourniquet was placed 10 cm below the fibular head with a pressure of 250 mm Hg. Following the surgical protocol, antibiotic prophylaxis with 2 g of intravenous cefazolin was administered 1 hour before surgery. Given the postaxial

Figure 5. BILATERAL POLYDACTYLY OF THE FIFTH TOE



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Figure 6. PREOPERATIVE RADIOGRAPH



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polydactyly, the lateral toe was to be excised;¹ a racket-shaped incision was made over the lateral supernumerary toe, and the sixth toe was completely removed from both feet.

A preplanned resection of the lateral head of the fifth metatarsal was performed using a microsagittal oscillating saw with a reshaping of the prominent bony edges. The authors consider this to be the most important surgical step because the proper functionality of the fifth radius in the long term depends on this action. Then 3-0 synthetic absorbable suture (Vicryl suture 3-0; Ethicon) was used to close the deep fascia and Biosyn 4/0 (Covidien) to close the skin with continuous suture. Wound closure was performed in planes and was finished with a sterile compressive dressing with a nonadherent absorbent dressing and the use of flat postsurgical shoes.

Postsurgical radiology was performed before the patient was discharged. During the immediate postoperative period, the patient was treated with a pharmacologic analgesic regimen (ibuprofen 600 mg/12 hours and paracetamol 500 mg/8 hours in case of pain), and the wound was inspected 72 hours after the operation during the first dressing change. The patient did not report pain at any time.

The skin closed by primary intention without modification. The external suture was removed after 10 days. No postoperative infection was recorded. Foot width, surgical scar, and radiology were evaluated at 3 months. After 6 months of follow-up, the patient was wearing normal shoes with no complaints or pain and was very satisfied with the aesthetic result of both scars and his feet in general (Figure 8). The patient was asymptomatic and reported no discomfort in daily life. His American

Orthopaedic Foot and Ankle Society Fifth Metatarsophalangeal Interphalangeal score improved from 48 to 62 (out of 100), accompanied by an improved numerical pain score compared with preoperative scores. At the end of a 1-year follow-up, a functional, painless, and aesthetically acceptable foot was observed. There was no neurologic dysfunction or movement restrictions noted in the operated feet.

DISCUSSION

Postaxial polydactyly is a malformation that occurs with or without duplication of the metatarsals or phalanges, with a wide clinical variability.²⁶ It can be treated conservatively with footwear modifications or more definitively with surgery; that said, clinical management guidelines for the condition are lacking. There is no consensus or description in the literature regarding the ideal age for surgical intervention. It is thought that the earlier it is treated, the better the outcome for the patient. However, given that the motivating factors for surgery are very specific, in some cases surgery is considered at an adult age.²⁰ In the illustrative case, surgery was performed in adulthood. Some providers favor surgery before the infant begins to walk to avoid alterations in function and gait biomechanics; others prefer delaying it until the bone maturation process in the affected radius is complete.²³ What most authors do agree on is individualized surgical treatment, pointing to decisive factors such as circulation, presence of hypoplasia, the state of fusion, alignment, and aesthetics.²⁷

In the literature, different surgical treatments for lateral and medial polydactyly are described. In 2021, Cheng et al¹⁶ reported a case of bilateral postaxial polydactyly associated with hallux valgus in both feet in a 22-year-old woman with the removal of the medial toe.

Figure 7. POSTOPERATIVE RADIOGRAPH



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Figure 8. FINAL WOUND HEALING WITH SATISFACTORY AESTHETIC SCAR



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Oragui et al²¹ described the case of a 17-year-old girl with an extrametatarsal head with a duplicated fusion of the proximal phalanx, and Han et al¹⁸ reported a case of polydactyly of the fourth metatarsal treated by bone transfer of the extrametatarsal bone and subsequent fixation with an internal plate. But polydactyly is not always visible, as reported in a case of polymetatarsia or metatarsal duplication associated with nonvisible polydactyly.²⁵

Most of the literature agrees that the most frequent type of polydactyly is unilateral postaxial polydactyly, except for those cases associated with a family history, which is usually bilateral. However, polydactyly is often coincident with other diseases or genetic syndromes. The Venn-Watson and Blauth-Olason classifications are the most comprehensive and encompass all the possible forms of presentation of the congenital anomaly.⁴

However, controversy in the literature persists in deciding the criteria for removing the excess toe. Surgical management is challenging, and there is no consensus for either preaxial or postaxial polydactyly. When the medial fifth and lateral sixth toes are of similar size, it is often difficult to decide which toe should be excised. In the present case, determining ray dominance was the most important factor for surgical planning. Some authors²¹ recommend removal of the most hypoplastic toe; others²² suggest the removal of the lateral digit because it is the least vascularized. Recently, Saijo et al²⁸ concluded that the lateral sixth toe excision for postaxial polydactyly of the foot produces good postoperative aesthetic outcomes. These authors point out the advantages of the lateral incision and the natural position of the reconstructed foot after surgery and do not report any major complications.²⁸

In 2018, Kubat and Antičević²⁰ reported in a retrospective cohort study that the medial ray was preserved, and the lateral one excised in most cases of postaxial polydactyly. The surgical procedure was decided at the discretion of the surgeon-in-charge through a preoperative

conference based on clinical presentation. In the present case, the guidelines for the treatment of polydactyly were followed, and the most lateral digit was excised. This technique requires a lot of skill and presents a steep learning curve. To avoid complications, it should be performed by a well-trained surgeon with in-depth knowledge of the principles of foot surgery.

In contrast, other authors¹⁷ suggest the removal of the medial toe because this maintains a larger interdigital space and avoids any loss of sensation of the external part of the foot, as well as adduction of the toe. In 2017, Escudero et al¹⁷ reported a case of postaxial polydactyly in a 52-year-old woman with the preservation of the supernumerary lateral toe with a plantar plate and medial collateral ligament reconstruction to improve forefoot biomechanics by stabilizing the metatarsophalangeal joint structures. Despite this study, there is little evidence on the stabilization of the metatarsophalangeal joint after amputation.

Researchers have demonstrated that treatment of foot polydactyly requires careful preoperative assessment, including radiographs and photography.²⁸ The results of this case showed that, postoperatively, the patient's gait improved, and the area of pain was eliminated. The patient had a substantially altered plantar pressure distribution with medial foot progression. The authors believe this treatment should improve forefoot biomechanics.

Further, a goal of surgery is to improve aesthetics, and a primary concern was the aesthetic aspect of the scar. Many factors influence patient satisfaction regarding their surgical scars: length, degree of hypertrophy, keloid formation, pigmentation, and discomfort. This patient described high satisfaction with his scars, given that they were on the lateral part of the foot.

CONCLUSIONS

The authors present a descriptive analysis of the surgical management of a Y-shaped metatarsal with biphalaengeal sixth toe and an accompanying review of the literature with an eye toward improving surgical treatment protocols from a clinical experience perspective. Bilateral postaxial polydactyly can be treated in adulthood with an adequate physical examination and radiology, resulting in a good prognosis. Surgical treatment of this malformation should be individualized to account for functional/biomechanical and aesthetic considerations. ●

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