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SPECIAL TOPIC

Hand

Surgical Reconstruction for the Triphalangeal Thumb

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Summary: The triphalangeal thumb poses a complex reconstructive challenge to the congenital hand surgeon due to its rarity and variable anatomy. We discuss the available evidence, reporting clinical characteristics and outcomes of surgical reconstructive procedures of triphalangeal thumb alongside a representative case. The congenital hand surgeon must approach each patient with triphalangeal thumb individually to optimize the use of available tissues to maximize functional and aesthetic outcomes. *(Plast Reconstr Surg Glob Open 2023; 11:e5379; doi: 10.1097/GOX.0000000000005379; Published online 3 November 2023.)*

INTRODUCTION

The triphalangeal thumb represents a complex reconstructive challenge for the congenital hand surgeon. These thumbs, with three phalanges instead of two, present within a remarkable spectrum of differences, making treatment an exercise in careful evaluation and tailored interventions. Further complicating the care is the rarity of this difference. Pre-axial polydactyly is a common congenital hand difference, whereas triphalangeal thumb and triphalangeal polydactyly are far less prevalent. The incidence is higher in Sweden, Australia, and the Netherlands, where it occurs with a frequency of one in 16,000 live births,[1] and lower in other parts of the world, with one in 25,000 being commonly cited as the prevalence in the literature.[2] Given the rarity, treatment algorithms differ and are based on limited case series with the highest impact publications coming from centers with higher prevalence.

Triphalangeal thumb may occur as an isolated finding, or in the setting of other limb differences. The term five-fingered hand describes the hand with an isolated triphalangeal digit lying in the same plane, along with absent thenar musculature, a narrow first-webspace, and a fully developed extra phalanx with no sesamoid bones. The triphalangeal digit lying outside the plane of the hand is considered a thumb, however hypoplastic or developed it may be. Triphalangeal thumb may present with preaxial or radial polydactyly, postaxial or ulnar polydactyly, and syndactyly of the ulnar digits or ulnar and/or radial duplicates.[3,4] It may be associated with syndromic presentations, lower limb differences, or an autosomal dominant inheritance pattern.[5] Familial cases, which are most common, are often associated with preaxial polydactyly and bilateral presentations.[6] Hovius et al noted that 62% of the triphalangeal thumb patients in an area with a high number of familial cases also had preaxial polydactyly, and two-thirds had bilateral differences.[2,7]

Early theories as to the origin of the triphalangeal thumb were based on clinical observations of the five-fingered hand, revealing features more consistent with index finger formation, such as distally located epiphysis on the metacarpal and typical fingerprint patterns.[8] Based on embryologic assessments, we now understand that sonic hedgehog (SHH) expression in the developing hand drives the differentiation of the radial digits, with the thumb being the last to develop.[9,10] Triphalangism can be induced in chick embryos by increasing the level of SHH.[11] This work demonstrated that preaxial polydactyly, radial hypoplasia, and ulnar dimelia all occur on a spectrum, depending on both the timing and strength of ectopic SHH exposure.[12] This work has been demonstrated clinically in familial pedigrees as well.[13,14] SHH acts on the Gli3 transcription factor, and this appears to be reduced in cases of triphalangeal thumb. Over-expression of HoxD12 is also associated with triphalangeal thumb formation.[15] Genetic testing in familial cases has mapped the pathogenic locus to be the zone of polarizing activity regulatory sequence. Polydactyly is seen more often in these zone of polarizing activity regulatory sequence-associated mutations, whereas hypoplastic variations are seen more often with mutations in growth and differentiation pathways.[16] Hypoplastic variations are more often seen with congenital malformations and in the setting of Holt-Oram syndrome, Blackfan-Diamond syndrome, and Fanconi anemia.[7,17,18]

With profound variability in clinical presentation, many authors have sought to delineate classification

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schemes, but without substantial success in attaining uniform use. Published classification systems focus on the clinical features of the triphalangeal thumb, such as the shape of the accessory phalanx,[20–22] opposition,[23–25] syndactyly and polydactyly,[7] and type of polydactyly present.[20,22,24,26,27] The complexity of deformity in triphalangeal polydactyly has made it difficult to place in the Wassel-Flatt categories. The triphalangeal thumb is considered a malformation under failure of axis formation/differentiation hand plate and under radial/ulnar axis in the Oberg Manske and Tonkin classification.[10,28] The most commonly used designations are the Wood's classification of the accessory phalanx and the ability of the triphalangeal digit to achieve opposition.

The surgeon treating patients with triphalangeal thumb should recognize the unique anatomy of the individual patient and tailor treatment accordingly. The surgeon should be mindful of thenar muscle deficiency or weakness, webspace contracture, and joint instability. Zuidam et al have shown that the collective effect of these differences results in decreased grip and opposition strength in both childhood and adulthood for all patients with triphalangeal thumb regardless of which type. They found, interestingly, that despite this functional limitation, untreated adult patients perceived fewer functional deficits and instead expressed greater dissatisfaction with the appearance of their thumb.[19,29] Indeed, case reports of incidental findings of triphalangism reinforce this finding.[30] We highlight familial triphalangeal thumb in twin pediatric patients to present the surgical decision-making involved to address triphalangeal thumb. We further review surgical management options for triphalangeal thumb and associated clinical characteristics.

CASE REPORT

Twin boys with bilateral triphalangeal thumb duplications (Wassel-Flatt type VII) presented with their mother and maternal grandfather, who both also had bilateral triphalangeal thumb duplications (Figs. 1 and 2). The mother had her duplicate removed in childhood, but the grandfather had not removed his and had found the additional digit to be helpful as a carpenter. The mother desired surgical reconstruction for the twin boys.

Given the polydactyly, surgical intervention was recommended at an early age to allow cortical patterning with a single thumb. The twins underwent staged surgical correction, with one hand at a time to limit functional deficit during surgical recovery. The first intervention involved removal of the radial duplicate and was performed at the age of 19–20 months. This was then repeated on the contralateral hand approximately 10 months later. Findings of note included a widened trapezium with a cup-in-cone carpometacarpal joint on the radial duplicate and a saddle carpometacarpal on the ulnar duplicate. The trapezium was reduced along with the removal of the radial duplicate, and extrinsic tendons and intrinsic musculature inserting on the radial duplicate were transferred to the retained ulnar digit.

Takeaways

Question: What are the options for surgical reconstruction of the triphalangeal thumb?

Findings: Triphalangeal thumb is a rare and complex reconstructive challenge for the congenital hand surgeon. Surgical options are described in the context of achieving the goals of reconstruction: reducing thumb length; maintaining stable joints; and improving position, function, and aesthetics. Surgical options include reduction osteotomy and fusion; excision with ligament reconstruction; and adjunctive procedures such as epiphysiodesis, pollicization, tendon transfer, Z-plasty, and skin flaps.

Meaning: Surgical correction of the triphalangeal thumb must be specific to the individual patient.

At age 4, one twin underwent revision on the right side to release a scar band and tighten the metacarpophalangeal joint (MCPJ) radially. MCPJ laxity can become more pronounced after removal of triphalangeal thumb with polydactyly due to intrinsic imbalance as the metacarpal grows. At age 10, he underwent reduction osteotomy at the distal interphalangeal joint (DIPJ) and epiphysiodesis of the thumb metacarpal on the one hand, and four months later, the procedure was repeated on the contralateral hand in conjunction with a Z-plasty deepening of the first webspace. The decision for epiphysiodesis was made at the point at which his metacarpal length was approximately the same as his father's. At 12 years old, his postoperative results demonstrated continued right hyperextension and MCPJ laxity (Fig. 3). Though not done in this patient, as his function was not limited by the laxity and he did not want to undergo further surgery, other options to address MCPJ hyperextensibility include volar plate plasty and sesamoidesis.

At age 4, the other twin underwent reduction osteotomy at the DIPJ on the one hand due to the presence of a delta phalanx and significant clinodactyly. At age 10, he underwent reduction osteotomy at the contralateral DIPJ, along with thumb metacarpal epiphysiodesis. At 12 years old, his postoperative results displayed adequate form and function, though also with some right thumb MCPJ laxity (Fig. 4).

DISCUSSION

Given the wide phenotypic variation of the triphalangeal thumb, surgical correction must be specific to the individual patient (Table 1). The ultimate goals are to reduce thumb length, improve position and function, and maintain stable joints. The group in the Netherlands has distilled its experience into an excellent algorithm to help guide congenital hand surgeons through some of the decision-making for these patients.[8]

Addressing the Middle Phalanx

The ideal length of the thumb places the tip of the digit at the level of the index PIPJ. In a triphalangeal thumb with

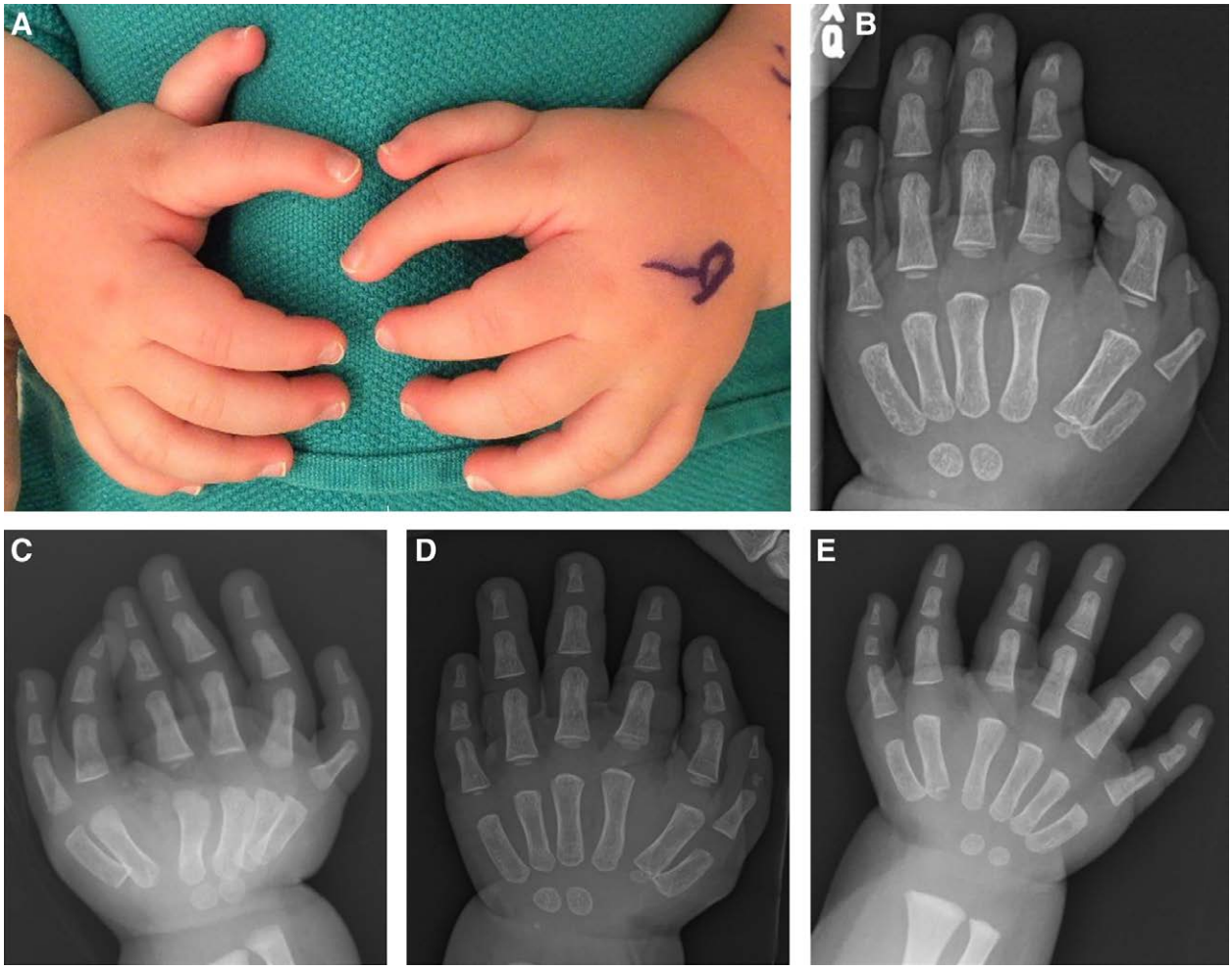

Fig. 1. Twin boy preoperative images and radiographic films. A, Clinical picture demonstrating bilateral triphalangeal thumbs of the first twin. B, Preoperative radiograph of the first twin's left hand. C, Preoperative radiograph of the first twin's right hand. D, Preoperative radiograph of the second twin's left hand. E, Preoperative radiograph of the second twin's right hand.

relatively normal motion and function, simple resection of the extra phalanx, or reduction osteotomy and fusion may be utilized to correct thumb length and appearance. In more complex cases, addressing the extra phalanx may be just one among several surgical interventions to achieve optimal aesthetic and functional outcomes. Earlier described procedures, such as amputation of the distal phalanx, result in inferior cosmesis. Concomitant reduction osteotomy and fusion are usually recommended in cases where the extra phalanx is large and well formed, whereas excision along with ligament reconstruction is recommended in cases where the extra phalanx is smaller and associated with clinodactyly.[31] Reduction osteotomy in both the longitudinal and transverse directions can be used to correct angulation and length.[32] This procedure's surgical approach may be made through a spiral incision to allow for skin resection as described by Girsch et al,[33] or through straight line incisions as described by Hovius et al.[7] In selecting which phalanx to which to fuse the added phalanx (proximal or distal), an assessment of range of motion along with clinical inspection for joint creases as an indicator of suitable joint function may guide the surgeon. Fusions to both the proximal and distal phalanx have been reported with good outcomes, although the distal joint is the most commonly selected joint for fusion.[33–36]

Resection of the accessory phalanx with ligament reconstruction was an early treatment, offered by Milch in 1951.[37] It has been reported to have a high rate of stiffness or late instability[22]; however, subsequent authors have reported many case series with overall satisfactory outcomes.[21,38,39] Girsch et al have suggested that joint stiffness may be present before surgery and should not be regarded as a complication.[33] Upton has suggested that results of this procedure are less predictable in children over the age of 4 years, and therefore encouraged reduction osteotomy instead for older patients.[40] Hovius et al found no outcome differences comparing a series of 16 thumbs treated with reduction osteotomy and fusion to 17 thumbs treated with excision and ligament reconstruction for patients with an average age of 3 years.[41] Resection of the accessory phalanx may be aided by Z-plasty release of the skin on the tight side and excision of skin on the long side in patients with deviated joints. Collateral ligament reconstruction can be achieved by shortening on the long

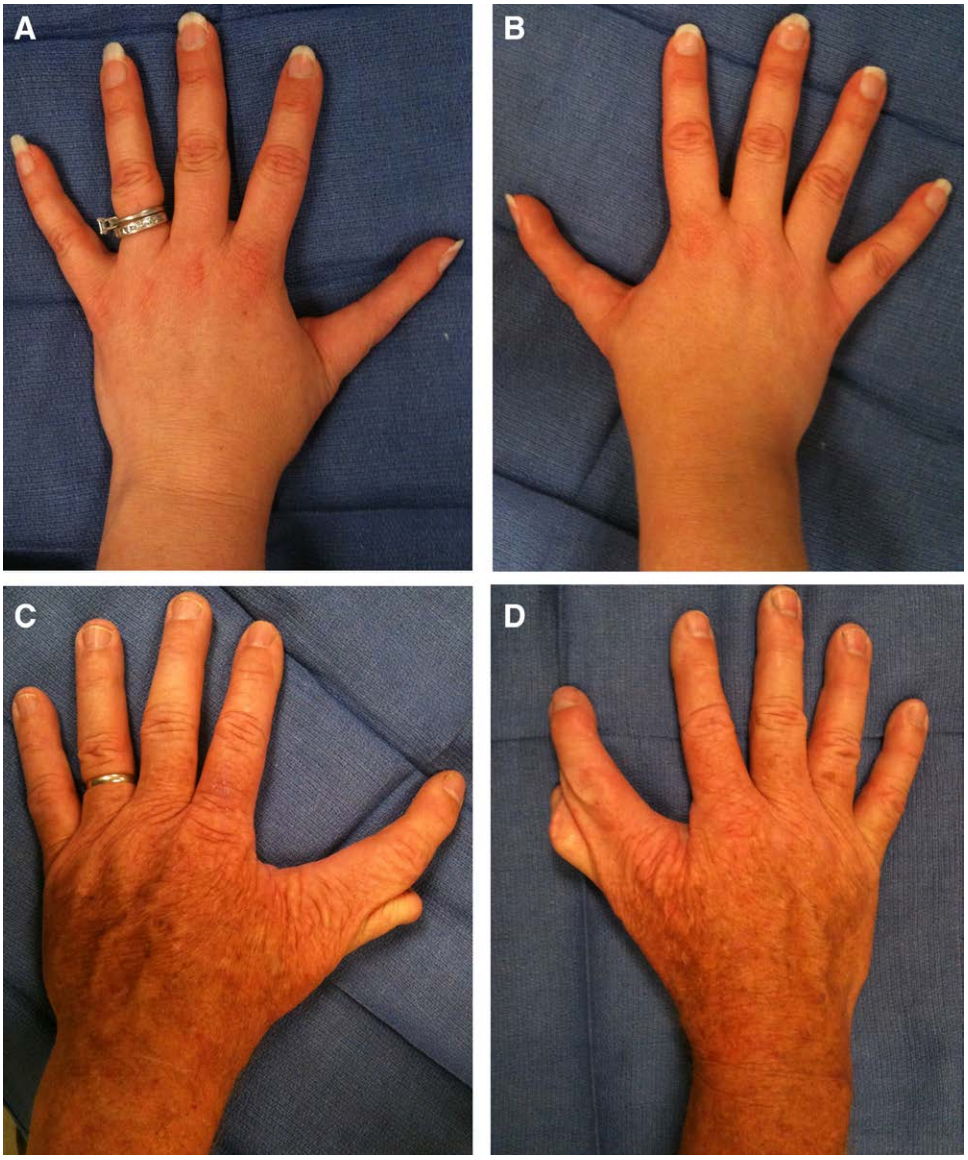

Fig. 2. Clinical pedigree of the twin boys with bilateral triphalangeal thumbs. A, Left hand of the patients' mother after thumb duplicate resection. B, Right hand of the patients' mother after thumb duplicate resection. C, Left hand of the patients' grandfather with untreated triphalangeal thumb. D, Right hand of the patients' grandfather with untreated triphalangeal thumb.

side.[7] V-to-Y lengthening of the collateral ligament on the short side can be used as an adjunct in patients for whom clinodactyly persists.[6] With no clear data to support one technique over the other, the decision to manage the phalanges with resection or osteotomy and fusion becomes a matter of preference, patient age, size and shape of the extra phalanx, and assessment of joint mobility before intervention.

Addressing the Metacarpal

The thumb length may additionally be impacted by an abnormally long metacarpal, making reduction of the extra phalanx by resection or osteotomy insufficient alone for restoration of optimal thumb length. The metacarpal epiphysis of the triphalangeal digit may be found proximally, as is normal for the thumb ray; distally, as is normal for the fingers; or at both ends of the metacarpal. This results in differing thumb growth depending on which variation is present. Zguricas et al demonstrated that the thumb metacarpal length in all patients with triphalangeal thumb is longer than the metacarpal length in the unaffected general population, and this difference is independent of the patient's presenting type of triphalangeal thumb.[42,43] The patients with the longest metacarpals relative to the general population tend to have the five-finger hand presentation and tend to have the epiphysis positioned distally, or a double epiphysis.[44] Zuidam et al recommended shortening procedures to be performed before bone growth completion, with the metacarpal shortened more for

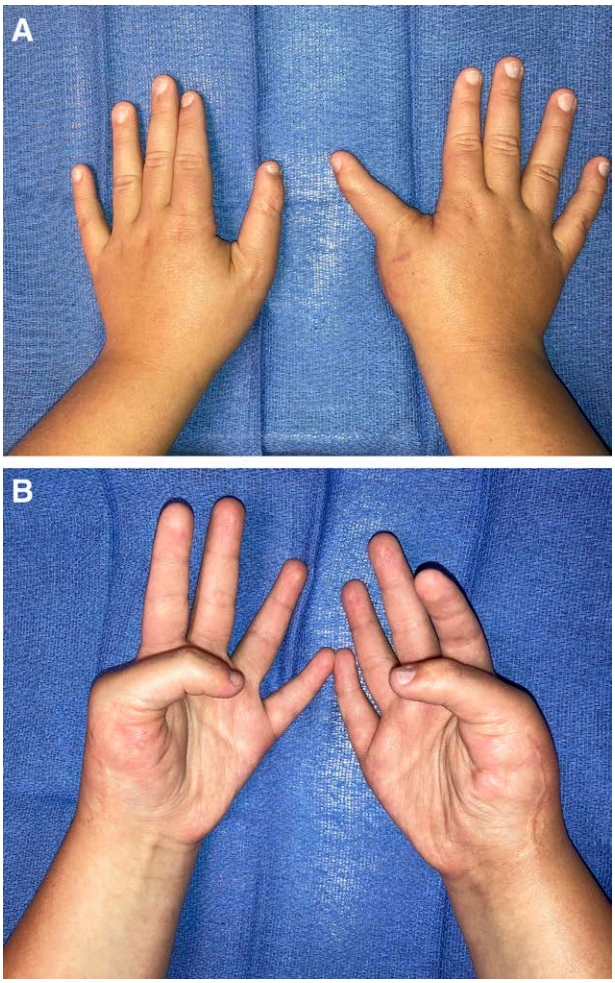

Fig. 3. Postoperative images after surgical reconstruction for twin A triphalangeal thumbs at age 12. A, Dorsal postoperative view. B, Volar postoperative view.

the patients with a double or distal epiphysis and just sufficiently for optimal thumb length relative to the index finger for patients with a proximal epiphysis.[42] Consideration could also be given to epiphysiodesis of the distal growth plate in patients with double epiphyseal plates to help minimize unpredictable growth of the digit.[45] El-Karef described his approach to treating excess metacarpal length. In the first of a two-stage procedure, he removed the extra phalanx as well as performed an oblique osteotomy to both shorten the metacarpal and place the thumb in a position of abduction and pronation. This was performed concurrently with fusion of the distal portion of the extra phalanx to the proximal portion of the proximal phalanx to remove the accessory phalanx. The second stage procedure was an opposition tendon transfer. He reported good outcomes for 15 thumbs treated in this fashion.[46] The rotation osteotomy of the metacarpal can additionally aid opposition by positioning the thumb and is recommended for all patients by Nieuwenhoven et al despite their observation that many of these patients develop hyperextension at the MCPJ secondarily.[7] The authors recommended concurrent shortening of the extensor tendon and intrinsic muscles, but not the flexors. Secondary corrections may be performed to address MCPJ hyperextension, including volar plate arthroplasty and sesamoidesis.[47,48] Hovius et al reported that approximately 9% of their patients required this procedure.[6]

Considering potentially needing to shorten the digit at both the phalangeal and metacarpal level, some authors have instead recommended pollicization of the long triphalangeal thumb, allowing the proximal phalanx to replace the long metacarpal, and substituting the MCPJ for the carpometacarpal joint (CMCJ).[20] Results published by the group in the Netherlands, however, suggest that pollicization produces results inferior to combination reduction osteotomy of the phalanx and metacarpal shortening.[7] In select patients with severely hypoplastic digits, however, pollicization may produce better functional outcomes.[6,49] Moreover, in patients with limited mobility of the CMCJ, a pollicization of the triphalangeal digit may improve motion by effective ablation of the stiff joint and transition of the MCPJ to act as the CMCJ.[46]

Restricted Webspace

A restricted webspace can also severely impact hand function. Surgical interventions described include Z-plasty releases, although in situations where an extra phalanx is being removed or shortened for fusion, redundant tissue may be advanced into the webspace. Local flaps from the dorsal hand and adjacent index finger have been described[22,50] along with larger local options like the radial forearm flap.[40] Advanced options include pedicled flaps raised on the digital vessels of resected duplicates.[51]

Considerations for Triphalangeal Thumb in Polydactyly

When dealing with a triphalangeal thumb and polydactyly, there is added difficulty when determining which digit to ablate and which to retain. Given the embryologic origin of the triphalangeal thumb, it is not uncommon to have a better digit in the ulnar position, as the collateral ligaments can be retained to help with joint stability. Patients may challenge the surgeon by presenting with a triphalangeal digit, which is more developed but with deficiencies in joint motion, and a biphalangeal digit that is hypoplastic but with better joints. The syndactyly and additional digits may further impair hand function by blocking the motion of the best thumb, making a clinical assessment difficult. Treatment options reported in the literature include ablation of the triphalangeal duplicate regardless of the functional status of the biphalangeal duplicate, modified pollicization, and vascularized transfer of the best joint into the retained digit.[52] Also reported is the on-top plasty, wherein a superior ulnar digit is transferred on top of the metacarpal to make the best of both parts. This can achieve excellent results for patients with a single well-formed CMCJ.[53] When two CMCJ are present, transfer to the radial metacarpal may risk CMCJ instability later, as the radial CMCJ is frequently less developed

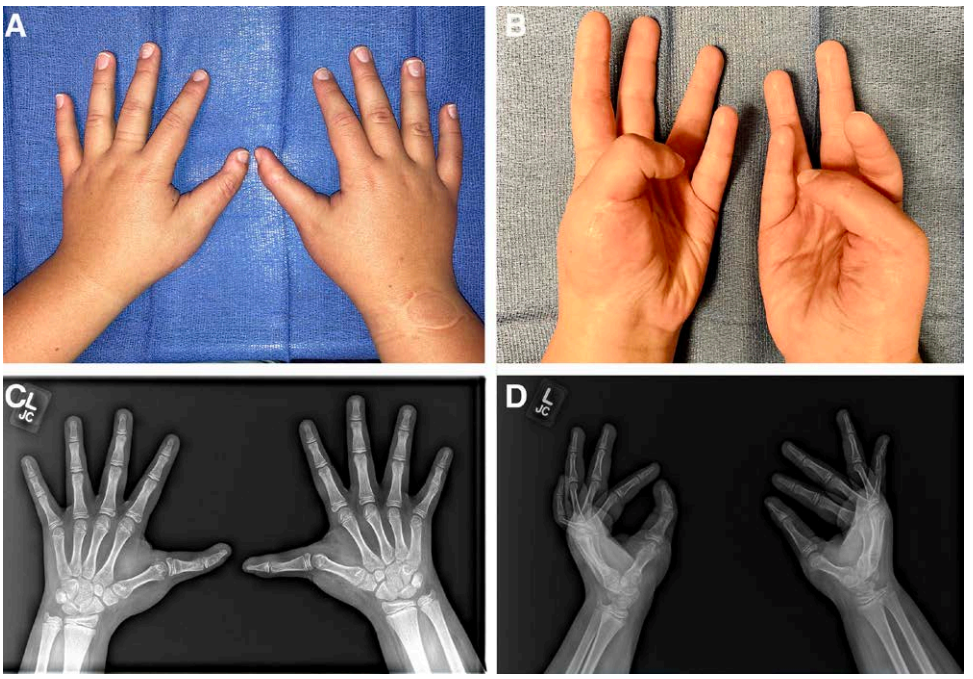

Fig. 4. Postoperative images after surgical reconstruction for twin B triphalangeal thumbs at age 12. A, Dorsal postoperative view. B, Volar postoperative view demonstrating opposition. C, Anteroposterior postoperative radiograph. D, Oblique postoperative radiograph.

than the ulnar CMCJ. After resection of the duplicate digit(s), patients still benefit from shortening osteotomies and supplemental procedures, as discussed above. The syndactyly and polydactyly may provide the surgeon with additional tissues for reconstruction, and the surgeon is encouraged to make use of all available parts by incorporating the additional skin into the webspace for deepening when possible. At the time of resection and reconstruction, care should be taken to ensure thenar musculature is appropriately reinserted and accessory extrinsic tendons are rebalanced as needed. Patients with polydactyly may also need nailbed narrowing and other tailored procedures.

Improving Opposition

Deficiency in opposition can be among the most functionally detrimental aspects of triphalangeal thumb. Children may have limitations in pinch, lateral pinch, and spherical and cylindrical grasp tasks,[46] although untreated adult patients do not seem to experience this as having an impact on their daily living.[29] Options for reconstruction include the standard opposition transfers with donors including the abductor digiti minimi, flexor carpi ulnaris, extensor indicis proprius, and flexor digitorum superficialis of the ring finger. Caution should be exercised when selecting the flexor digitorum superficialis transfers as patients with a weak CMCJ are at risk for metacarpal subluxation.[6] Opposition transfers are offered as a secondary reconstruction only for patients that require them, and in the experience of Nieuwenhoven et al, only 20% of their patients required a tendon transfer for opposition.[7]

Surgical Timing

With regard to timing of surgical intervention, individuals with nonopposable presentations cannot perform pinch activity and are likely to benefit from intervention early in childhood,[44] although waiting until just before school entrance at 3 or 4 years of age to perform osteotomies is reasonable.[7] Doing so has the added advantage of allowing the epiphyseal plates to be more clearly seen on plain films. Patients with polydactyly may benefit from earlier intervention to remove the extra digits and allow for cortical patterning with the single thumb, with recommendations for surgery between 1 and 2 years of age.[33] Observation of the child in play may help guide the surgeon, as those children who are using the thumb to pinch and pick up objects may be able to delay intervention until closer to school age, whereas those using the index and middle finger for pinch should be operated on sooner to allow them to adapt to using the reconstructed thumb.[6] Given the advice of Upton, early surgical intervention would be appropriate in children who are candidates for accessory phalanx excision.[40] Also benefiting from earlier intervention may be those patients who have significant clinodactyly from an abnormally shaped extra phalanx.

CONCLUSIONS

Triphalangeal thumb represents a broad spectrum of clinical presentation, with no unified classification scheme or treatment algorithm. The congenital hand surgeon must carefully evaluate individual patients and work with the family to select a treatment that best addresses

Table 1. Patient Individual Factors Guiding Surgical Management of Triphalangeal Thumb

Structure	How It Can Present Individually	Available Treatment Options
Thenar muscle	Opposition absent or deficient	• Opponensplasty (avoid FDS if CMC unstable) • Pollicization of the radial most digit • Ablation of a hypoplastic radial digit and pollicization of a functionally superior finger • Rotation osteotomy to a more functional position
First webspace	Contracture	• 4 flap Z-plasty • Adjacent tissue transfer from resected ray if polydactyly • Adjacent tissue transfer from shortening during fusion or resection of additional phalanx • Local flaps • Distant pedicled flaps
Middle phalanx	• Delta • Rectangular • Fully developed	• Amputation of the tip of the thumb—not recommended • Resection of the accessory phalanx with ligament reconstruction • Reduction osteotomy—requires at least one functioning joint to be preserved and a relatively larger additional phalanx
MCP joint	Hyperextensible	Volar plate plasty and sesamoidesis
Metacarpal	• Longer than normal • Epiphysis proximal • Epiphysis distal • Epiphysis both ends • In plane of the hand • Out of plane of the hand	• Shortening and repositioning into abduction and pronation • Epiphysiodesis • Pollicization
CMC joint	• Stiff • Unstable	• Consideration for pollicization for function • Reduction of trapezium if widened
Carpus (especially scaphoid and trapezium	• Hypoplastic • Malformed • Absent	• See CMC, no specific reconstruction
Polydactyly	• Biphalangeal • Triphalangeal • Hypoplastic • Well-formed • Multiple	• Nailbed narrowing • Pollicization • On-top plasty • Ablation of hypoplastic digit • Combination of best parts

both function and aesthetic concerns with careful consideration of making the best use of the available tissues to maximize outcomes.

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DISCLOSURE

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