The Duplicated Thumb: A Review

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Abstract

Keywords

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- preaxial polydactyly
- Bilhaut-Cloquet procedure
- ablation
- reconstruction

Preaxial polydactyly is a common, often sporadic, congenital anomaly that must be addressed surgically early in life. Ideally, the surgeon seeks to accomplish three goals: construction of a thumb that is adequate in size, preservation of pinch function, and reconstruction of all components in one procedure. Although each case is unique, several classification systems attempt to describe the various types.

In this article, the authors discuss the various classification systems, procedures, and outcomes after surgery for pediatric thumb duplication.

Epidemiology

Congenital deformities of the hand are common, affecting at least 2.3 per 1,000 total births.¹ Thumb polydactyly is the most prevalent duplication in the Caucasian and Asian populations, occurring in approximately 0.8 to 1.4 per 1,000 births.^{2–4} This particular hand condition is most often a cosmetic rather than functional issue, as the supernumerary thumb often contains all of the sensory and motor units that are necessary for function. Moreover, the duplicated thumb often works in concert with the whole hand as a contributing unit.⁵ Most individuals with thumb duplication will undergo excision of the extra thumb, however, for both functional and cosmetic reasons.

Thumb duplications result from a failure of the radial-ulnar axis of the hand plate to form and/or differentiate. The zone of polarizing activity (ZPA) in the posterior part of the developing limb bud is the principal signaling center. The ZPA expresses sonic hedgehog protein, which controls the formation of radial-ulnar features. Abnormal expression of Hox genes, bone morphogenic protein, and the Gli-3 gene all play a role in the evolution of thumb duplications.⁶ Genetic consultation is not indicated, as most cases are sporadic and unilateral. However, the presence of a triphalangeal thumb is known to exhibit an autosomal dominant inheritance pattern.^{4,7}

Classification

Thumb duplications are classified under the more commonly used Oberg, Manske, and Tonkin (OMT) classification as malformations, because of their failure of formation/differentiation of the hand plate in the radial-ulnar axis.⁸ The deformity itself can be further subdivided based on the specific form of duplication. The Wassel system, developed in 1969, is most familiar among pediatric hand surgeons for its simplicity and is the most widely used system in the recent literature (►Table 1).^{9,10} This scheme describes seven types of radial polydactyly according to the level of the skeleton at which duplication occurs.⁶ Types I to IV classify distal-to-proximal levels of thumb duplication, and type VII describes radial polydactyly with a triphalangeal component (►Fig. 1). Type IV is the most common type of thumb polydactyly, followed by type II and type VII (►Fig. 2).^{11,12} Despite its widespread use, however, this system has several limitations when it comes to surgical planning. For example, the Wassel classification often fails to identify the origin of the extra digit, a key morphologic characteristic necessary for the process of surgical planning. Additionally, the system does not align classification types with unique operative techniques; hence, the management of Wassel types II, IV, and VI may be entirely similar.¹³ In clinical practice, this terminology is best used as a basic framework for the management of a thumb duplication.⁶

Table 1 Wassel classification of thumb polydactyly

Type I	Bifid distal phalanx
Type II	Duplicated distal phalanx
Type III	Bifid proximal phalanx
Type IV	Duplicated proximal phalanx
Type V	Bifid metacarpal
Type VI	Duplicated metacarpal
Type VII	Triphalangeal

Others have attempted to improve upon the Wassel system, including Buck-Gramcko, Upton, and Flatt.^{14–16} The more recent Rotterdam classification, introduced in 2008 by Zuidam et al, consists of eight types of radial polydactyly and adds letters to describe aberrant components, followed by their location. By combining elements of the Wassel, Buck-Gramcko, and Upton taxonomies, it accounts for joint involvement and associated anomalies in all radial polydactylies, including the triphalangeal thumb and thumb triplication.^{17,18} A study by Dijkman et al compared the reliability of the Wassel and Rotterdam classifications.¹⁹ Out of a study population of 520 cases, only 60% could be classified using the Wassel classification, compared with 100% using the Rotterdam classification. The unclassifiable cases under the Wassel system had aberrant components, mostly of the triphalangeal, deviating, and hypoplastic kind, which called for specific surgical plans. Drawbacks to the Rotterdam classification include its complexity and the lack of visibility of certain aberrant components on radiography at a young age.¹⁹

Preoperative Assessment

A detailed history and physical examination is important in all of these patients. It is important to pay particularly close attention to thumb motion and functional use, and status of the thenar musculature and first webspace. The thenar musculature can vary from normal to hypoplastic in Wassel types IV–VII, while the first webspace may be narrow in more proximal duplications (Wassel types IV–VIII).⁶

Mobility is largely determined by the integrity of the carpometacarpal (CMC) joint. The CMC joint, which plays a larger role in thumb mobility than the metacarpophalangeal (MCP) and interphalangeal (IP) joints, is typically normal in more distal duplications, but may be mal- or underdeveloped in proximal duplications. Joints distal to the duplication may also be hypoplastic, even though the better-developed thumb may exhibit near-normal movement.¹⁷ Children have more difficulty with tip pinch with loss of flexion at the IP joint, but they often compensate well enough to preserve functionality. Stability of the distal thumb is more important than motion, and if the CMC joint is intact, motion should be sacrificed for stability if necessary.⁶

While observing appearance and movement, the examiner should become familiar with tendon location and functionality. A flexor pollicis longus (FPL) and extensor pollicis longus (EPL) may be present in both thumbs, although the more functional thumb usually demonstrates better tendon quality and active range of motion (AROM). These tendons may also have eccentric insertions, which can cause distal convergence of the duplicated thumbs.¹⁷ Lastly, the shape of the thumb must be considered, as appearance is of utmost importance to

Fig. 1 (A) Wassel Type II duplication, dorsal view, (B) Wassel Type II duplication, palmar view, (C) X-ray showing duplicated distal phalanx.

Fig. 2 (A) Wassel Type IV duplication, dorsal view. (B) Wassel type IV duplication, volar view. (C) X-ray showing duplication of proximal and distal phalanges.

the patient.⁶ The duplicated thumbs usually retain smaller nails and pulp compared with the unaffected contralateral side, although the better-developed thumb typically arises from the ulnar side and is the larger of the two in the majority of cases. Finally, the affected hand should also always be compared with the unaffected side when possible,¹⁷ as bilateral involvement should alert the examiner to a possible syndrome, such as Townes-Brocks syndrome.²⁰

Radiography is necessary to provide information about the pattern of bone involvement and monitor changes over time. This is typically performed around one year of age, or at the time of surgery. X-rays performed prior to one year can be less helpful, as there is a lack of ossification of the hand skeletal system.²¹

Surgical Treatment

Considerations

All duplicated thumbs require surgical intervention, as the potential for improved function and appearance outweighs the risks of surgery. In all cases, a stable, mobile thumb of appropriate size and shape is the goal. To achieve this, surgical correction must address more than just bony structural problems. Regardless of type, reconstruction must also address musculotendinous abnormalities, including collateral ligaments and tendons for appropriate mechanical stability.¹⁸ Simple ablative surgery alone in these cases can result in instability and malalignment of the remaining thumb.⁴

Although the timing of surgery is somewhat arbitrary, reconstruction is typically performed as pinch function is developing, between 1 and 2 years of age, and before the development of fine motor skills.¹⁷ The main goals of reconstruction are an improved appearance, and the restoration and maintenance of function and pinch activity.²⁰

When faced with a patient with radial polydactyly, hand surgeons specialized in congenital anomalies must analyze the preoperative functional potential and degree of hypoplasia of both thumbs to decide the best operative technique. Subtle type I or type II thumbs with a common nail and proper anatomical alignment may not require surgery. Ablation and reconstruction can be used for all Wassel types; however, type I is more difficult. The Bilhaut-Cloquet procedure, which involves removing the central portion of the nail and soft tissues of each thumb and combining these down the middle, has fallen into disfavor due to consistently poor cosmetic results. It is usually better to attempt to salvage most of one thumb with its nail intact and utilize a portion of the other to “fill out” the remaining thumb.

In all cases, as supernumerary thumb elements are resected, autologous tissue becomes available for grafting. This additional tissue may be incorporated for mechanical support or as appropriate for transpositional techniques. Soft tissue flaps derived from the resected thumb can also augment the radial side of the retained thumb to make it more cosmetically comparable to the uninvolved side. The “on-top plasty” (transfer of the better-developed distal tissues of one thumb onto the better-developed proximal tissues of the other) has also been used in certain cases. Finally, in severe cases where both duplicated thumbs are hypoplastic or with significant triphalangeal cases, the most appropriate approach may well be pollicization. It is important to remember and also remind parents that multiple surgeries may be necessary to achieve a satisfactory result, and the thumb may never achieve optimal function. One study determined the frequency of different methods used for treating duplicated thumbs and found that the overwhelming majority of cases were treated with resection and reconstruction (85%). Simple

ablation alone (5%), the Bilhaut-Cloquet procedure (8%), pollicization (1%), and the on-top plasty (1%) are used much less frequently.¹⁷

Ablation of the Radial Thumb

The most commonly performed surgery is ablation and reconstruction.²² This procedure involves removal of the smaller, usually radial, thumb elements with transfer of the collateral ligament and centralization of the extensor tendon. When thumb components are equal, the ulnar thumb is preferentially retained. This helps preserve the ulnar collateral ligament (UCL), an essential contributor to pinch function.

The radial thumb is incised using a dorsal incision to expose both of the EPL tendons. A portion of the radial thumb is often left as a flap to inset into the remaining thumb, particularly if the girth of the remaining thumb will be less than that of the opposite, normal thumb. These incisions are usually made in an interdigitating zig-zag fashion to avoid linear scars and later scar contracture (►Fig. 3). If the patient has an angular deformity of the IP joint, the initial exposure should additionally include the distal insertion sites of the FPL and EPL tendons. The extensor tendon to the lesser component is excised along with its respective digit. The remaining EPL tendon is centralized along the retained thumb. Centralization can be achieved with simple folding toward the midline or with distal detachment and reattachment to the midline.^{2,23} If exposure of the flexor tendon is sufficient, eccentric flexor tendons may be centralized in a similar fashion. If the remaining FPL or EPL tendons appear hypoplastic, tendon grafts from the resected tissue can be used for additional structural support.

The next step in reconstruction involves transposition of the radial collateral ligament (RCL). The exact dissection depends on the Wassel type, but ultimately involves sharp elevation of proximally based ligamentous flaps. For type I and type II thumbs, the RCL is elevated along with an osteoperiosteal sleeve off of the lesser cortical shaft. The width of this ligamentous/osteoperiosteal flap is determined by the anteroposterior diameter of the bone shaft. Elevation should continue proximally to a point that will later allow for osteotomy of redundant bone tissue. Type III thumbs typically

allow for an osteotomy design that preserves the proximal phalanx base and avoids the need to raise a ligamentous flap altogether. In type IV thumbs, the most common type, proximal exposure must be extended beyond the MCP joint. Type IV thumbs often will have thenar muscle attachments, such as that of the abductor pollicis brevis (APB), to the lesser thumb. The involved thenar muscles should be included in a musculoligamentous flap with the RCL. After proximal exposure is complete, a proximally based flap consisting of the periosteum, joint capsule, RCL, and the distal insertion of involved thenar muscles is elevated off of the lesser thumb component. Altogether, these elements are often termed the collateral ligament complex (CLC) and their reattachment to the base of the remaining phalanx is essential for the future stability of the MCP joint.

After raising the corresponding RCL flaps, attention is turned to neurovascular bundle dissection, FPL and EPL tendon tenotomy, and resection of the radial thumb. Type IV thumbs may contain further musculotendinous anomalies that should be addressed as well. Thenar muscles and extrinsic tendons may demonstrate anomalous attachments that cause deleterious mechanical imbalances if not corrected. For example, some thumbs may exhibit an abnormal connection between the FPL and EPL tendons on the radial side of the thumb, a structure termed the pollex abductus, which should be released to avoid a postoperative angular deformity.²⁴ Type IV thumbs may also exhibit a “U ligament” between the proximal phalanges of each thumb. This ligament may be dissected from its attachment to the lesser thumb and serve as an extra support for later RCL reconstruction.²⁵

Joint Reconstruction

After removal of the supernumerary thumb elements, proper longitudinal alignment of the thumb follows. In type I or II thumbs, abnormalities of the IP joint may be seen. Type IV thumbs require detailed inspection of both the IP and MCP joints. Deformities may include angulation of the joint (“zig-zag deformity”) or redundant proximal bone tissue that supported the previously resected thumb components. This extra tissue can be removed sharply or with a rongeur.^{4,14,26} A sharp scalpel blade may be used for removal of this extra bone

Fig. 3 (A) Wassel II duplication showing markings for excision and reconstruction. (B) Thumb after resection of radial digit and reconstruction. Note interdigitating flaps rather than straight-line scar.

in younger children prior to significant ossification. The excision of excess proximal digit or metacarpal bone is performed along its radial aspect with care not to disrupt the RCL. Further extra-articular malalignments may require additional wedge osteotomy with a scalpel or osteotome.

After adequate osteotomy, the previously raised RCL flaps are inserted onto the cartilaginous base of the retained digit using periosteal sutures.²⁷ The CLC flap is inserted into the radiopalmar aspect of the proximal phalanx. Radial thenar musculature and the reinsertion of the APB provides substantial stabilization to the reconstructed MCP joint.²⁸ Additional autologous reinforcement of the CLC using EPL or FPL grafts may improve long-term deformity and instability.⁸ The thumb is stabilized using a percutaneous Kirschner (K) wire across the joint.

Bilhaut-Cloquet Procedure

When thumb duplication is symmetric, with no identifiable lesser component, the Bilhaut-Cloquet procedure may be performed to retain elements of both thumbs. This procedure is technically challenging and often associated with poor outcomes such as nail splitting.²² It is typically reserved for type I thumbs only, and involves wedge resection of the central bone and nail, with approximation of the remaining lateral components. Approximation can be stabilized with either transverse K wires or sutures. This procedure has been plagued by long-term complications from imprecise matching of components that are required for normal digit and nail growth. Disruption of the nail fold often results in a split nail or a longitudinal ridge, whereas damage to the growth plate results in long-term deformities. For this reason, the epiphysis is typically left out of the wedge resection.

To avoid these complications, the Bilhaut-Cloquet procedure has been modified into an extra-articular technique that avoids operating across the IP joint.²⁹ This technique preserves a single physis from one thumb and uses the other thumb to contribute part of the distal phalanx for stability.

Wassel Type V, VI, or VII Reconstruction

Higher Wassel classifications follow the same principles of lower class thumbs, but may require additional soft tissue reconstruction. Narrowing of the thumb-index finger web space may require deepening with a Z-plasty technique or dorsal advancement flap. Intrinsic muscles that involve the lesser thumb must be resected and reattached to the remaining thumb.

Triphalangeal thumbs present the problem of an additional joint space. The number of joint spaces can be reduced through excision of the extra phalanx or arthrodesis. Arthrodesis is often the preferred technique and is typically performed across the joint with the least amount of motion.^{30,31} Many triphalangeal thumbs will not require any type of surgery as they can be quite functional, however.

More complex cases require great surgical ingenuity. As the anatomy becomes more complicated, the surgical approach becomes more individualized and typically involves trying to incorporate the best phalangeal portions of both thumbs into a single best thumb. On-top plasty is a specific example that combines the better-developed distal component of one

thumb with the superior proximal aspect of the other.¹³ When surgical reconstruction of the thumb components would otherwise fail to result in a stable, functional digit, the resection of both with pollicization of the index finger may be the best option.⁶

Postoperatively, the patient is placed in a long-arm thumb spica cast, which can be removed along with the K wires at 4 to 5 weeks postoperation.²² At this point, the patient can usually be allowed to move; however, some suggest that the reconstructed thumb be protected in a removable short arm thumb spica splint for an additional 4 to 8 weeks. This type of splinting in children can be difficult as they will usually not wear the splint as prescribed.

Outcomes

Postoperative results depend primarily on the preoperative diagnosis. For example, AROM, stability, alignment, and strength are more likely to be limited postoperatively in the radial polydactyly subtypes that require joint manipulation. The preoperative absence of an IP joint flexion crease indicates that the joint has never been flexed, and bodes poorly for future motion regardless of tendon reconstruction.² Other factors that affect postoperative outcome include the degree of angulation, hypoplasticity, and the presence of triphalangeal components.^{3,32,33} Those duplicated thumbs that are essentially straight and parallel ("balanced") will generally have a good outcome from reconstruction, whereas those that are angulated toward each other ("imbalanced") generally will have poorer outcomes (→Fig. 4).

Outcomes based on the surgical technique can vary significantly depending on several factors, including the appropriateness of the technique for the specific form of duplication. When used appropriately, simple ablation yields excellent results, and most cases will not require revision.⁶ On the other hand, the resection and reconstruction technique produces smaller, less stable thumbs with preserved IP and MP joint AROM.¹⁷ It is important to note that the scars associated with this procedure can be conspicuous (→Fig. 5). The Bilhaut-Cloquet procedure, comparatively, produces broader, more stable thumbs, but with diminished IP and MP joint AROM.

Fig. 4 "Unbalanced" Wassel type IV duplication. These can be difficult to reconstruct due to the angulation and instability of both interphalangeal joints.

Fig. 5 Thumb one year after reconstruction of type IV duplication.

The resultant scars of the thumb dorsum and thumb pulp are also often readily apparent with this procedure, and the resulting broad split in the nail has made this procedure unpopular.^{5,34,35} On-top plasty, performed in 1%, generally yields a thinner thumb with a stiff IP joint.¹⁷ When the UCL at the MCP joint requires repair, Hogan et al found that interference knot fixation of the tendon graft yielded superior results among four methods of reconstruction, demonstrating further that surgical technique plays an important role in functional as well as aesthetic outcome.³⁶

In 1983, Tada et al reported the functional outcomes of 237 radial polydactyly cases.⁴ The study found 76% good, 20% fair, and 4% poor results in patients undergoing initial surgical reconstruction with an average 3-year follow-up.⁴ Another study by Stutz et al followed patients for an average of almost 17 years after surgery and found similar outcomes, with 79% of patients with good and 21% with fair outcomes, suggesting that outcomes are well maintained over time.¹⁴ Despite these reported results, revisions are often required; studies have reported reoperation rates varying between 12 to 26% based on average follow-up ranging from 4 years to 22 years. In general, the reoperation rate increased the longer these patients were followed.^{3,14,37}

Several factors must be taken into consideration when evaluating thumb appearance postoperatively, including length, girth, nail width, and angulation. Although it was previously reported that the reconstructed thumb is typically diminished in terms of length compared with the normal thumb,^{2,38} Goldfarb et al found no significant difference with regards to these factors in their study of 31 reconstructed thumbs in 26 children with radial polydactyly.³⁹ It is interesting to note that girth did not differ significantly from the contralateral side, even though only four Wassel type I, II, or III thumbs were treated with a Bilhaut-Cloquet procedure. Relative nail width, however, was significantly decreased after surgery. The discrepancy between girth and nail width may have been secondary to the addition of skin and soft

Fig. 6 (A, B) Hands of child 6 years status post bilateral Wassel type IV duplication reconstruction. Thumbs are stable and functional. Note mild hypoplasia and somewhat prominent metacarpophalangeal joint due to broad metacarpal head. (C) Postoperative X-ray showing reasonable alignment. Again note somewhat broad metacarpal head.

tissue during reconstruction. The study also found that patients were more aesthetically displeased with IP joint angulation compared with MCP joint angulation after surgery. Patients with a reconstructed Wassel type VII thumb scored the lowest on subjective ratings of appearance. These results can aid the physician in developing family understanding prior to surgery (►Fig. 6).³⁹

Conclusion

The treatment of radial polydactyly is complex, as the clinical presentation is highly diverse, and each subtype requires a specific surgical approach. Surgeons must take into account the unique anatomy of each case before deciding on the appropriate operative technique and determining the best overall result. Outcomes vary and depend largely on the preoperative diagnosis and procedure performed. Despite this, treatment is met with generally good outcomes, which are well maintained over time.

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