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Review

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Current concepts in the management of the duplicated thumb

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Abstract

Preaxial polydactyly is a common congenital anomaly of the hand presenting at birth. Surgical treatment is aimed at creating a functional thumb capable of normal grip and pinch strength with acceptable aesthetics. Each case is unique and presents individual challenges to the hand surgeon. The aim of this review is to provide a synopsis of current knowledge and recommended surgical techniques for the duplicated thumb.

Keywords: Preaxial polydactyly, hand deformities, reconstructive surgical procedures, thumb duplication, congenital anomaly

INTRODUCTION

Preaxial polydactyly (also known as duplicated thumb, radial polydactyly, or split thumb) is a common congenital anomaly of the hand characterized by a duplicated, or partially split, first digit. The reported incidence is 0.08 to 1.4 in 1,000 live births and is highest in Caucasian and Asian populations[1]. The etiology of this malformation involves aberrant genetic signaling during development and thus its presentation at birth can be highly variable. Duplicated elements of the thumb can include phalanges and/or metacarpals and management depends on the exact type of duplication. Biphalangeal thumb duplication is most often unilateral and sporadic, but the triphalangeal thumb is more commonly symmetric, bilateral, and inherited in an autosomal dominant pattern[2,3]. Duplicated thumbs are rarely associated with other syndromes, and

therefore, it is not usually indicated to obtain further workup or genetic testing solely based on the presence of a duplicated thumb[4].

The origin of the duplicated thumb begins in the 4th week of embryologic development as the limb buds begin to form. The Sonic hedgehog (SHH) signaling pathway, a product of the zone of polarizing activity (ZPA) in the posterior mesoderm, regulates the development of the limb bud in the anterior-posterior axis. Disruption of this pathway, specifically the presence of ectopic SHH in the anterior mesoderm, results in preaxial polydactyly[5]. The severity of disruption has been shown to correlate with the severity of duplication; minor increased expression of SHH in the anterior mesoderm results in a biphalangeal thumb, while moderately increased expression of SHH results in a triphalangeal thumb[6]. Given that the SHH signaling pathway is imperative in the development of the upper extremity, disruption of this mechanism has been implicated in other clinically distinct congenital anomalies, including radial aplasia and ulnar dimelia[2,7].

ANATOMY & CLASSIFICATION

The duplicated thumb is a direct result of the failure of formation/differentiation and is thus classified as a malformation under the Oberg, Manske, and Tonkin (OMT) classification[8]. Further identification of a patient's specific type of thumb duplication is crucial for surgical planning. Several classification systems exist for thumb duplication, and each presents its own benefits and limitations. The most widely used is the Wassel classification published in 1969. This system describes anomalies based on the level of skeletal duplication[9]. Seven types of duplication are ordered distally to proximally, with type VII describing a triphalangeal thumb [Figure 1]. Type IV, representing a duplicated proximal and distal phalanx with a single metacarpal, is the most common form of duplication, with an incidence of approximately 40% [Figure 2]. Following this, Type II thumb is common, with an incidence of approximately 20%[4] [Figure 3].

Modifications of this system have been developed to account for further variations such as triphalangeal components or triplication, most notably the Rotterdam classification system. It achieves a higher level of specificity, but it is not as widely used[10,11]. Chung *et al.* expanded on other faults of the Wassel classification, including its poor applicability to surgical planning. Chung presented another classification based on the anatomic morphology at the origin of the extra digit[12]. Although these deficiencies exist in the Wassel classification system, its simplicity offers great reliability and clarity among surgeons and in the literature[13].

Adding to the complexity is the presence of abnormalities in the soft tissue that may be concurrent with the osseous abnormalities. Joint instability, aberrant insertions of the thumb musculature, abnormalities in the A1 and A2 pulleys, differences in nail morphology, and narrowed first web space are all soft tissue anomalies that may be present and must be accounted for in surgical planning[14].

PREOPERATIVE EVALUATION

The clinical presentation of a duplicated thumb is most often obvious and identified shortly after birth. An in-depth history and physical exam are important as each Wassel type presents its own challenges. The exam should include a careful assessment of overall appearance, differences in the surrounding musculature and first web space, and functional ability of the thumb. The two thumbs may be symmetric, or one may seem dominant over the other. However, both thumbs are often hypoplastic or underdeveloped to some degree compared to the normal contralateral thumb. Most often, the ulnar thumb is more developed and larger[15] [Figure 4]. Duplicated thumbs usually have a smaller nail than the contralateral side, but distal duplications (Wassel type I and II) may have broad nail plates which may be the only physical sign of duplication. Lastly, a thorough family history and evaluation for the presence of other congenital anomalies

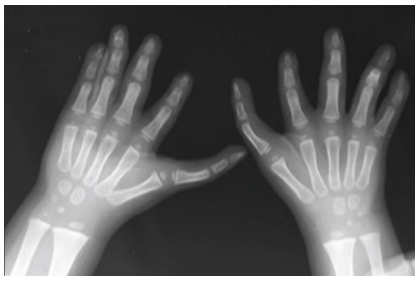

Figure 1. X-ray showing bilateral triphalangeal thumbs in a child.

Figure 2. X-ray showing a Wassel type IV thumb duplication.

Figure 3. X-ray showing a Wassel type II thumb duplication.

should be done, as it could point to an underlying syndrome[16].

A detailed understanding of normal anatomy is necessary for identification of abnormalities. Hypoplastic or malpositioned musculature and tendinous structures may influence the appearance and mobility of the thumb. Often, the ulnar thumb metacarpal is ulnarly deviated, while the proximal phalanx is radially deviated due to disrupted alignment of extrinsic tendons[17]. Thenar musculature may also be hypoplastic in Wassel type IV - VI (as well as in Wassel type VII with a triphalangeal ulnar thumb) and contribute to a shortened first web space[18].

Figure 4. Thumb of child with Wassel type IV duplication, note that the ulnar thumb is somewhat larger than the radial.

Thumb mobility at the carpometacarpal (CMC), metacarpophalangeal (MCP), and interphalangeal (IP) joints should be assessed. CMC joint mobility is crucial to the function of the thumb and is often preserved in distal duplications but can be abnormal in Wassel types V and VI. The stability of the CMC joint is prioritized over the MCP and IP joints, as pinch strength is compromised in the latter, but the functionality of the entire thumb is compromised in the former[18].

Radiographs are necessary to classify and evaluate the osseus abnormality. However, a major limitation of plain films is the inability to visualize unossified bone sites and cartilaginous structures of the infant hand. Du *et al.* proposed the routine use of three-dimensional spoiled gradient recalled echo (3D-SPGR) MRI in cases of bifurcation at the MCP joint level to evaluate the range of articular abnormalities that may be present but do not appear on radiograph[19]. Three-dimensional ultrasound is also an option to evaluate soft tissue anomalies, but neither ultrasound nor MRI replaces the need for radiography[20].

SURGICAL MANAGEMENT

The aim of surgical treatment of a duplicated thumb is that of any other reconstructive procedure: re-establishing form and function. To achieve this, one must create a functional thumb for optimal grip and pinch and achieve an appropriate size and shape for improved aesthetic appearance.

The timing of surgical intervention involves two opposing considerations. Reconstruction before the development of fine motor skills is important, but allowing enough time for the digits to grow will decrease the technical difficulty of the operation. Considering these factors, the generally accepted time to operate is between 1 and 2 years old[7,21,22]. Ultimately, this decision is up to the surgeon's discretion while taking into account the family's preferences. If the duplicate is interfering with the development of hand function, earlier reconstruction should be considered. It is also important to counsel the family on possible outcomes and, if necessary, temper expectations. It should be explained to parents that surgical treatment cannot create a completely normal thumb in form or function, but the goal should be to achieve significant improvement in both.

Ideal options for reconstruction are based on the preoperative evaluation and pattern of duplication [Table 1]. In mild type I or II duplications, no treatment may be necessary. When treatment is indicated, surgical options can be categorized into the following: (1) resection of the hypoplastic thumb; (2) resection with reconstruction of other elements including collateral ligaments or tendons; (3) the "on-top" plasty; and (4) the Bilhaut-Cloquet procedure. This procedure for a type I or II duplication is rarely utilized anymore,

Table 1. Wassel classification and associated options for reconstruction

Wassel classification	Options for reconstruction
Type I	No treatment Bilhaut-Cloquet Procedure Resection with Reconstruction
Type II	No treatment Bilhaut-Cloquet Procedure Resection with Reconstruction
Type III	Resection with Reconstruction
Type IV	Resection with Reconstruction
Type V	Resection with Reconstruction On-Top Plasty
Type VI	Resection with Reconstruction On-Top Plasty
Type VII	Resection with Reconstruction On-Top Plasty

as it leads to significant scarring and deformity of the thumbnail. The indication for each approach does not tend to correlate with a particular Wassel classification. For example, resection with varying degrees of reconstruction is commonly used for all Wassel types. This option is also by far the most performed procedure at a rate of 85%[15].

Resection of the hypoplastic thumb

As previously mentioned, distal duplications may require minimal or no surgical intervention. Simple excision can be performed for an obviously hypoplastic or floating thumb. For a skin bridge of less than 4 mm, Tonkin advocates for ligation in the neonatal period. In this procedure, the infant is held while a 27-gauge needle is used to infiltrate local anesthetic. A vascular clip or suture is then placed around the base of the hypoplastic thumb as close to its origin as possible and the digit is excised using a blade. The suture or clip is left in place to fall off on its own after approximately 2 weeks. For a skin bridge of more than 4 mm, surgical excision using the operating room and general anesthesia after 3 months of age is preferable[18]. Both options have been shown to result in good functional outcomes with satisfactory aesthetic results, but surgical excision showed lower rates of residual tissue and revisionary surgery compared to suture ligation[23].

Resection with reconstruction

Resection of the hypoplastic thumb followed by reconstruction is the most commonly performed operation[15]. However, this option encompasses a wide range of complexity depending on the bony and soft tissue elements that require reconstruction. Excision of the more hypoplastic (usually radial) thumb is performed, but this alone would result in an unstable thumb. Therefore, reconstruction of the collateral ligaments and flexor/extensor tendons is necessary to restore thumb stability for pinch and prehensile strength. If both thumbs are symmetric, the radial thumb should still be excised to preserve the ulnar collateral ligament because it is important for pinch function[16,24,25].

A zig-zag incision is made to minimize the risk of contracture and to ensure adequate soft tissue coverage [Figure 5]. A racquet incision is preferred by some, but the linear closure may contribute to contracture or growth limitation[18]. In the senior author's experience, a linear scar results in a good aesthetic result without any limiting contracture and is the preferred incision design. The distal insertions of the extensor pollicis longus (EPL) and flexor pollicis longus (FPL) tendons should be exposed if there is angular deformity of the IP joints. The incision should also proceed proximally as needed for exposure of the most proximally involved segment and to facilitate tendinous/ligamentous reconstruction. However, for more distal

Figure 5. One option for incisions to reconstruct a duplicated thumb, interdigitating flaps.

duplications (Wassel types I - III), reconstruction of the radial collateral ligament (RCL) may be simplified by designing the osteotomy distal to the base of the proximal phalanx and thus avoiding the need for dissection of the RCL.

Wassel types IV and above necessitate dissection of a proximally based flap that includes the RCL and thenar musculature to facilitate its reattachment to the retained thumb. As described by Manske, this dissection should take place in a plane deep to the tendons with an osteo-periosteal sleeve, taking care to preserve the origin of the RCL from the metacarpal[26]. In a Wassel type IV duplication, the abductor pollicis brevis (APB) may be attached to the radial thumb and should be included in the raised flap. The digital neurovascular bundles should be identified and protected during the dissection, although this may be complicated as the number and location of neurovascular structures is highly variable. The nerves should be dissected to their bifurcation before excision of the nerve to the radial thumb to avoid injuring the nerve to the retained ulnar thumb[27]. With the periosteal flap created, the radial thumb can now be excised along with the nail and nail fold. The skin of the radial thumb can be preserved and used as a neurovascular island flap for coverage of any deficiency of the reconstructed thumb. Additionally, the adipofascial tissues from the resected thumb may also be maintained to augment the girth of the preserved thumb. However, in the senior author's experience, using this excess tissue does not result in an improved cosmetic result [Figure 6].

Attention is then turned to joint reconstruction and corrective osteotomies. The IP and MCP joints should be closely inspected for separate facets, redundant bone, or angular deformities. A sharp scalpel blade, rongeur, or osteotome can be used to excise the redundant bone of the proximal phalanx and metacarpal. A percutaneous Kirschner wire is driven retrograde to stabilize the reconstructed joints.

The RCL is then secured to the reconstructed thumb using periosteal sutures. The remaining extensor and flexor tendons may need to be centralized on the retained thumb to rebalance and correct any angular deformity. This can be accomplished by folding the tendon towards the midline or detaching and reattaching it distally in a more anatomic insertion point. Another method of tendon rebalancing involves reattaching the tendons to the ulnar side of the proximal phalanx, as the tendons of the retained thumb tend to be abnormally inserted on the radial side of the retained thumb's base of the distal phalanx[28]. If the excised thumb's tendons are removed, they can be used as an autologous tendon graft for additional support of the RCL, reconstruction of the pulley system, or for severely hypoplastic EPL or FPL tendons of the reconstructed thumb[29]. Lastly, Type V or VI duplications may also have a shortened first webspace that should be addressed with a Z-plasty or dorsal advancement transposition flap[24].

Figure 6. View of thumb after reconstruction with the utilization of some soft tissue from the excised digit. Note the thickened, uneven appearance of the reconstructed thumb.

The on-top plasty

In rare instances, neither thumb can be chosen for reconstruction. With more proximal duplications, complex anatomy may show a superior distal nail and pulp on one thumb while the other contains better proximal components. The on-top plasty, created by Soilan in 1961, was originally used for traumatic amputations but later applied to preaxial polydactyly cases in which both thumbs contain inadequate proximal or distal components[30,31,32]. In this procedure, osteotomies of both digits are performed and the distal parts of one thumb are transposed on a neurovascular pedicle to the proximal parts of the other to fashion a more functional singular thumb[33] [Figure 7].

The Bilhaut-Cloquet procedure and its modifications

Symmetric thumb duplication also poses increased difficulty of reconstruction. The Bilhaut-Cloquet procedure, which has been shown to have a high rate of complications along with a poor aesthetic result, may be chosen in this instance to retain elements of both thumbs. Often used for Type I or II thumbs, the procedure involves wedge resection of the bone and nail from the central portion of both thumbs. If the nail width of one of the thumbs is 70% of the contralateral normal thumb, it is recommended to retain that nail in its entirety with the excision of the other duplicated thumb's nail. Otherwise, both nails are divided and combined[34]. The bone is then secured using suture, interosseus wires, or Kirschner wires. Precise approximation of the two physes is imperative to ensure that thumb growth is not disturbed. To avoid this complication entirely, the epiphysis is often excluded from the wedge resection[24]. This approach is generally to be avoided, and it is best to save the best thumb and discard the other.

Modification of the procedure has been used by avoiding the IP joint with an extra-articular osteotomy, retaining the epiphysis of only one phalanx while the other phalanx contributes to the distal portion[35].

POSTOPERATIVE CONSIDERATIONS

After reconstruction, a short or long-arm thumb spica cast is used for 4-5 weeks. K wires are removed after radiologic assessment of bony union, usually around the time of cast removal. An orthoplastic thumb spica splint may be used at night for an additional 4-8 weeks if further protection of the reconstruction is needed[24]. Formal therapy for the reconstructed thumb is not usually indicated, as the child will naturally be mobile. However, scar massage and passive motion are important components of postoperative healing in which parents can participate[36].

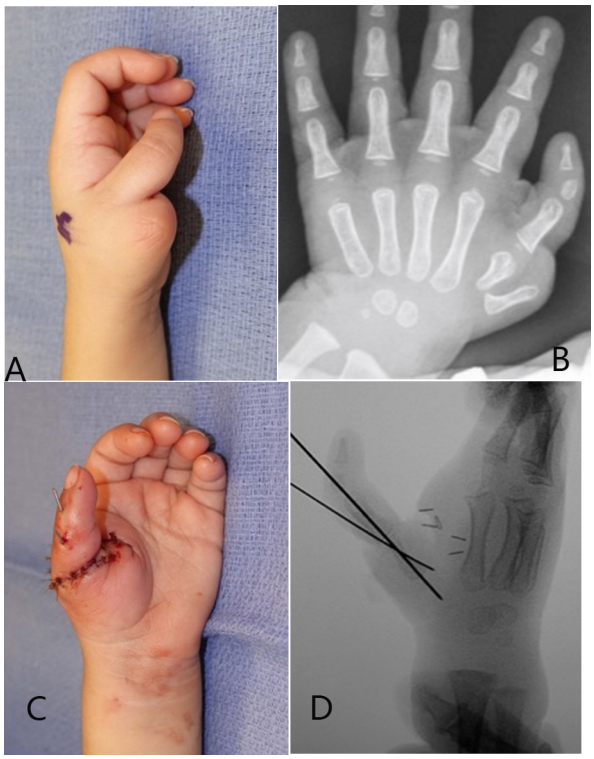

Figure 7. (A) Preop view of duplicated thumb. The radial thumb is attached to the metacarpal and shortened, while the ulnar thumb is essentially "floating"; (B) Preop X-ray showing bony structure; (C) View after transfer of ulnar thumb "on top" of radial thumb; (D) X-ray showing fixation of ulnar thumb on radial thumb.

OUTCOMES

Overall, reconstruction of the duplicated thumb has been shown to be successful in creating a long-term functional and aesthetically acceptable digit[37]. However, postoperative complications and revisional surgery occur with cases involving higher complexity as determined in the preoperative evaluation. Increased degrees of angulation, triphalangeal components, hypoplasticity, and decreased motion preoperatively all contribute to increased difficulty of reconstruction and, therefore, increased risk of poor outcomes[16]. Ogino *et al.* also found increased complications in Wassel types III, VI, and VII[33]. Age greater than 3 years old at the time of reconstruction is also correlated with worse outcomes[38].

The most common postoperative complication is axis deviation[39]. Other complications include joint instability, scar contracture, and poor aesthetic result. The on-top plasty has shown good results in long-term sensation, grip and pinch strength, and union of the osteotomy[30]. However, the Bilhaut-Cloquet procedure's technical difficulty often results in worse outcomes that include nail dystrophy, disruption of normal growth of the digit, and IP joint stiffness[35].

CONCLUSION

The duplicated thumb remains a challenging, nuanced area of congenital hand surgery. A new understanding of its epidemiology, especially related to other congenital anomalies, is still in progress. Additionally, its management is difficult to condense into an algorithm. Successful outcomes are heavily reliant on the expertise of the surgeon and adequate preoperative planning. Using the principles as outlined above, outcomes are generally good and can provide a long-term functional thumb for the child throughout their life.

DECLARATIONS

Authors' contributions

Conceptual design of the manuscript: Gimenez A
Drafting of the manuscript: Moreno T, Gimenez A
Critical review and editing of the manuscript: Gimenez A, Pederson WC

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Conflicts of interest

All authors declared that there are no conflicts of interest.

Ethical approval and consent to participate

Patient consent was obtained for the use of photographs in this study.

Consent for publication

Written informed consent for the publication of the images has been obtained.

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