



Congenital bifid thumb correction: causes of reoperation

Correzione congenita del pollice bifido: cause di rioperazione

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Summary

The aim of this study is to report results and complications occurred after surgery for congenital bifid thumb. From June 2015 to June 2020 a case series of 47 thumb duplications, Wessel type II, III and IV, was treated. All patients received removal and reconstruction of the thumb. They were evaluated using Tada score and plain X-ray with a mean follow-up of 6 years. Average Tada score was 3.8. Nine patients developed minor complications. A second surgery was necessary in 7 patients: 2 scar revisions, 3 corrective osteotomies, and 3 extensor pollicis longus tendon (EPL) retensionings. The percentage of reoperated patients is comparable with the data reported in the literature. Complications are minor and affect soft tissues. This is due to the difficult management of soft tissues and their possible evolution. EPL lag is difficult to treat and in our experience it has a tendency to recurrence after retensioning. Level of evidence: II.

Key words: congenital bifid thumb, reconstruction

Riassunto

Lo scopo di questo studio è quello di riportare i risultati e le complicazioni si sono verificati dopo l'intervento chirurgico per il pollice bifido congenito. Da giugno 2015 a giugno 2020 è stata trattata una serie di casi di 47 duplicati del pollice, Wessel tipo II, III e IV. Tutti i pazienti hanno ricevuto la rimozione e la ricostruzione del pollice. Essi sono stati valutati utilizzando Tada score e pianura raggi X con un follow-up medio di 6 anni. Tada punteggio medio era 3.8. Nove pazienti hanno sviluppato complicazioni minori. Un secondo intervento chirurgico è stato necessario in 7 pazienti: 2 revisioni della cicatrice, 3 osteotomie correttive, e 3 sondaggi estensori lungo tendine (EPL) ritensioni. La percentuale di pazienti recidivati è paragonabile ai dati riportati in letteratura. Le complicanze sono minori e colpiscono i tessuti molli. Ciò è dovuto alla difficile gestione dei tessuti molli e alla loro possibile evoluzione. Il ritardo di EPL è difficile da trattare e nella nostra esperienza ha una tendenza alla ricorrenza dopo il retensioning. Livello di prova: II.

Parole chiave: pollice bifido congenito, ricostruzione

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Conflitto di interessi

Gli Autori dichiarano di non avere alcun conflitto di interesse con l'argomento trattato nell'articolo.

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Introduction

Thumb duplication is a preaxial polydactyly with an incidence of 1:3,000 newborns, representing 6.6 % of hand deformities ^{1,2}. Typically, thumb polydactyly

occurs sporadically and unilaterally, but it may also be associated with Holt-Oram syndrome, Fanconi anemia, or Rubinstein-Taybi syndrome, thus requiring a thorough record of patient's medical history and physical examination with attention to other congenital abnormalities. Holt-Oram syndrome affects 1 in 100,000 births and is characterized by a variety of cardiac defects and hand anomalies, with thumb hypoplasia being the most common. Typically, patients with Holt-Oram syndrome have been diagnosed before examination by a hand surgeon. Fanconi anemia is a rare condition affecting 1 in 300,000 births and is characterized by bone marrow failure. In one series, 1 of 83 patients with preaxial polydactyly had Fanconi anemia. Rubinstein-Taybi syndrome affects 1 in 125,000 births and is characterized by growth delay, dysmorphic facies, intellectual disability, and duplication of the distal phalanges of the thumb. These conditions are rare, therefore they are not routinely screened unless the related medical signs can be observed³⁻⁶.

Thumb duplication is recognised among "duplications" in the International Federation of Societies for Surgery of the Hand (IFSSH)/Swanson classification⁷, among "abnormal formation/differentiation of the hand plate in the radio-ulnar axis" in the modified Oberg-Manske-Tonkin (OMT) classification^{8,9}, and among "late limb patterning/differentiation in abnormal axis differentiation in hand plate" in the latest update of the OMT classification¹⁰. According to the recent literature, its pathogenesis seems to involve Sonic Hedgehog Homolog protein (SHH), a signaling molecule produced in the Zone of Polarizing Activity (ZPA), a mesenchymal area containing signals that lead the limb to grow along the anteroposterior axis in vertebrates⁸.

The Wassel morphological classification is the most commonly used to guide the surgeon in decision making¹¹. It includes 7 types of deformity based on the level of the duplication. Wassel type IV thumb is the most common variant and occurs in 40% of patients, while type II thumb is the second most common variant and occurs in approximately 20% of patients. Although this classification is widely used for its ease of use, it shows a limited correlation with postoperative clinical results¹². Typically, bifid thumbs are underdeveloped and have varying degrees of abnormal tendons, bones, ligaments, and vasculature. To integrate the Wassel classification, several subsequent modifications of the original Wassel classification have been proposed, including triphalangeal subtypes, type IV subtypes, and systems incorporating symphalangism, deviation and triplication¹³⁻¹⁵.

Radiographs should be obtained to correctly assign classification and guide the surgical planning. Fingernail involvement in Wassel type I or II deformity requires a different surgical planning compared to type III or IV deformity. Preoperatively, it is important to emphasize to parents that, although surgical reconstruction improves appearance and

function, the operated thumb will never be identical to the unaffected contralateral, with the preoperative joint angulation, stability, and motion providing a clue to the resulting thumb function¹⁶.

Although little evidence is available regarding the most suitable timing for surgery, most authors recommend reconstruction between 1 and 2 years of age. This facilitates skeletal growth, it allows a less technically challenging reconstruction and the development of fine motor skills, such as thumb-index tip (pincer) grasp. Despite this, surgery can be safely delayed until after 2 years of age for patients with multiple comorbidities or when a more complex reconstruction is required. Historically, simple excision of the less-developed digit was the treatment of choice until follow-up revealed unsatisfactory functional and cosmetic outcomes. Specifically, excisional management alone frequently resulted in joint instability and off-axis or absent pull of the tendons on the preserved MCP and IP joints. Excision without reconstruction frequently results in a Z deformity of the residual thumb, which does not provide proper collateral ligament support and in-line pull of the flexor and extensor tendons. Increasing emphasis has been placed on thorough reconstruction of all abnormal structures in the retained thumb because of these unsatisfactory outcomes^{1,4,17-20}.

The main goal of the surgical correction is the creation of a stable, well aligned, functional and cosmetic thumb. Treatment of radial polydactyly is complex, as the clinical presentation is highly diverse, with each subtype requiring a specific surgical approach. Surgeons must take into account the unique anatomy of each case before deciding on the most suitable correction technique for the best overall result. The outcomes vary and depend largely on the preoperative diagnosis and procedure performed. Tonkin and Al-Qattan reviewed and described the most commonly used methods of surgical correction. In general, a correct surgical treatment of DT gives good well-maintained outcomes^{21,22}.

The aim of the study is to report the authors' case series between 2008 and 2013, considering all the causes that led to resurgery in order to improve both functional and cosmetic impairments of the thumb.

Materials and methods

Between June 2015 and June 2020, 47 thumb duplications in 41 patients (26 males and 15 females) were treated at the Authors' Institution. Mean age at first surgery was 16.4 months (range 11-26). 36 caucasian patients and 5 african patients were treated. According to the Wassel classification¹¹, 28 patients had type IV duplication, 10 type III, 7 type II and 2 type I. All the patients were treated by two senior surgeons with removal of the radial supernumerary thumb

and reconstruction of soft tissues when required. 30 cases had retensioning of the radial collateral ligament (RCL) at the MCP joint, 8 cases had rebalancing of the extensor pollicis longus (EPL) tendon, 8 cases had subtraction osteotomy of the proximal phalanx or the first metacarpal bone and 2 cases had additive osteotomy with a bone graft from the donor finger in MP clinodactyly. Postoperative treatment included arthrorisis with Kirschner wire for 5 weeks with a thermoplastic static splint to maintain the thumb in abduction and opposition. The splint was worn night and day for the first 2 months and during the night for another 2 months. Patients were periodically evaluated at the Authors' Paediatric Hand Surgery Outpatients Clinic. Mean follow-up was 6 years (range 3-8 y). Thumb function was evaluated for each patient using the Tada score²³, in association with plain X-rays (antero-posterior and lateral).

Surgical technique

The procedure is usually performed using dorsal zig-zag incisions, followed by deep dissection and inspection of the joints. Slight traction is established to enable evaluation of the IP or MCP joint congruency. Mainly in types II and IV, chondroplasty of the radial facet of the proximal phalanx and the metacarpus is necessary to give congruency to the joints, then followed by their testing for stability. In all the cases involving radial resection, the radial collateral ligaments are reattached with preservation of the capsular-periosteal flaps. Soft tissue from the base of the proximal phalanx is sutured to the metacarpal neck to reconstruct the MCP joint. The thenar muscles and the radial collateral capsular-periosteal flap are reattached at the same site. At this point of the procedure, if some joint instability remains, part of the EPL or flexor pollicis longus (FPL) tendon is used as a circumferential pulley around the proximal phalanx, suturing the tendon to itself. If there are connections between the FPL and the extensor tendons, they may be cut out. In order to ensure joint axial alignment, it is important to avoid eccentric pull of the EPL tendon by its radial insertion on the base of the distal phalanx. The duplicated EPL tendon of the dissected thumb, if present, may be sutured to its corresponding remaining tendon of the ulnar thumb at the ulnar base of the distal phalanx. The eccentric position of the tendon is then modified, and its deforming forces are consequently neutralized. At this stage, the soft tissue procedures described above allow proper alignment in nearly all the cases. If good alignment is not achieved with these procedures, osteotomy becomes necessary. Ulnar-base head-neck junction osteotomy of the proximal phalanx or of the metacarpal bone may be necessary to achieve axial alignment in some cases, mainly in types II

and IV, respectively. Corrective osteotomy is controversial in the youngest patients. When osteotomy is indicated, a 0.7-mm K-wire is introduced from the tip to the base of the thumb and left in place for 4 to 6 weeks. Any delta phalanx needs to be removed and corrective osteotomy is indicated. Z-plasty of the first web space is performed mainly in type IV thumbs when the first-space contracture limits their functionality. An elasticizing silicone cream is recommended starting from week one after suture removal to reduce retracting scars.

Results

All 41 operated patients (100%) were included; 7 of them were treated bilaterally. Clinical evaluation with Tada score²³ revealed a good or satisfactory functional result in 90.8% of patients, with a mean score of 3.8 points (range 1-5) and a mean follow-up of 6 years. In case of reoperated patients, the Tada score obtained before the second procedure was reported. Plain radiographs showed a good realignment of the thumb axis in 19 cases (86.3%), clinodactyly in 2 cases and Z deformity of the thumb in one case. Nine out of 47 thumbs (19%) developed minor isolated or associated complications: 4 hypertrophic surgical scars, 3 EPL deficits, 2 clinodactylies for MP and IP instability respectively, 1 nail dystrophy and 1 Z deformity. Seven patients (14%) were reoperated to treat complications on average 22 months after the first procedure (12-38 months): 2 scar revisions with Z-plasty, 2 wedge osteotomies in subtraction; 1 corrective double osteotomy of the metacarpal and the first phalanx for Z deformity; 3 EPL retensioning for active extensor lag of the distal phalanx.

Discussion

Polydactyly is one of the most frequently observed congenital hand malformations. Being such a common upper limb malformation, investigation into its cause and the consequent therapeutic measures is highly relevant. Various studies suggest that a gene localized to chromosome 7q36 is responsible for preaxial polydactyly types II and III. The Wassel classification is the universal classification for thumb duplication. From the time it was first described to the present day, triphalangism has been progressively considered as a different condition to that of thumb duplication, since it requires a different approach²⁴. According to many authors, the Wassel classification should only be used as a preliminary evaluation, since it does not consider the anatomical alterations of the soft tissues and skeletal dysmorphisms such as thumb triplication, triphalangism, thumb hypoplasia, deviation and symphalangism, as reported in the classification proposed by Zuidam and colleagues

in 2008¹⁵. These anatomical variations of the thumb, if incorrectly treated, can lead to postoperative complications along the whole first ray²⁵. A careful intraoperative evaluation is therefore mandatory in order to plan the appropriate correction of the soft tissues in order to limit the incidence of complications.

The surgical procedure has four well-defined aims for the dominant thumb: functional alignment, stability, overall good function and satisfactory cosmetic appearance. A decrease in motion range is acceptable to achieve IP and MCP joint stability. Historically, types II and III thumbs with symmetrical radial and longitudinal sizes have been treated with the Bilhaut-Cloquet procedure²⁶, although this procedure is currently under discussion because of some concerns regarding split nail deformity, stiffness, and late deformity caused by epiphyseal damage and growth arrest. Recently, some authors also reported a modified Bilhaut-Cloquet technique that offers satisfactory results with radial resection and soft tissue reconstruction adopted in Wassel type IV thumbs with divergent MCP and convergent IP joints^{22,24,27}.

The removal of one of the two thumbs is indicated in type III and higher and in cases of a remarkable asymmetry of the two thumbs in type I and II. This technique includes the removal of the hypoplastic thumb, usually the radial one, in order to preserve the ulnar collateral ligament (UCL) at the IP or the MP joint, in association with the reconstruction of

the ligamentous and tendinous complex or osteotomy^{22,28}. The complications reported in the literature include excessive joint instability and Z deformity²⁵.



Figure 1. Most frequent causes of reoperation: articular instability (Wassel type II-IV). a) Thumb Z deformity; b) scar hypertrophy; c) extensor deficiency (EPL); d) nail-dystrophy (Wassel type I-II).

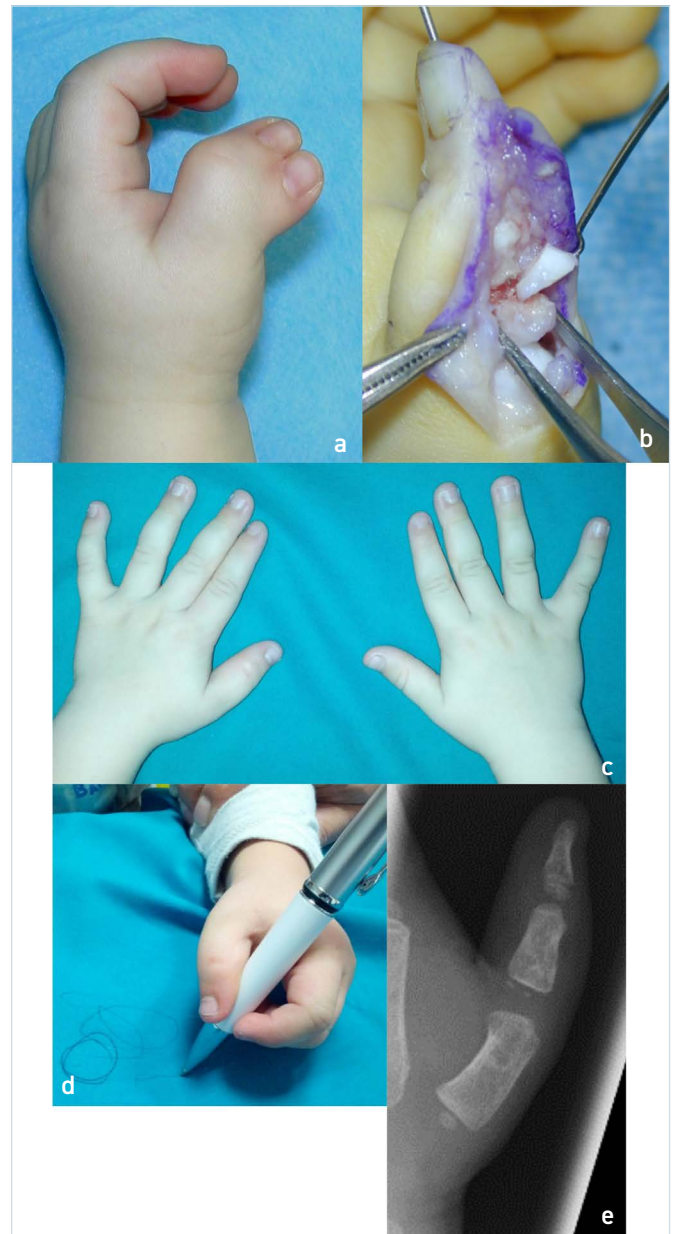


Figure 2. Patient with type IV deformity (Wassel classification), (a-b) preoperative image, operated at 12 months with removal of the radial thumb, in association with additive osteotomy with graft of donor thumb, RCL reconstruction and EPL retensioning (c-d). Clinical and radiological evaluation at 30 months post-op, showing restoration of the anatomical axis with optimal clinical and functional results (Tada score 5) (e-f-g).

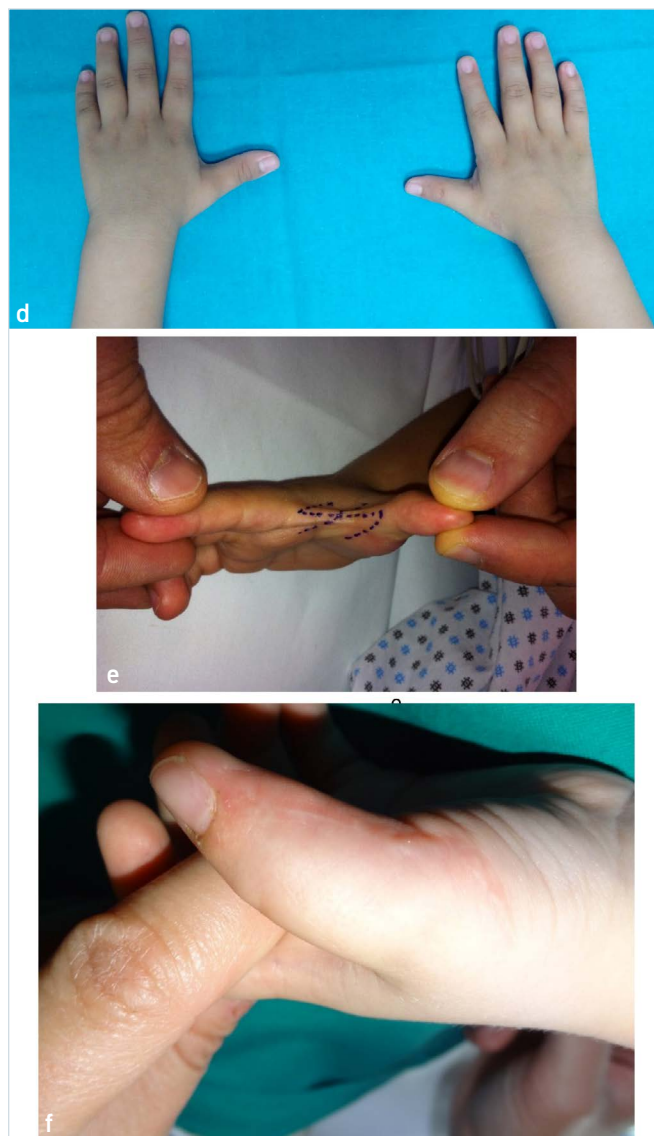
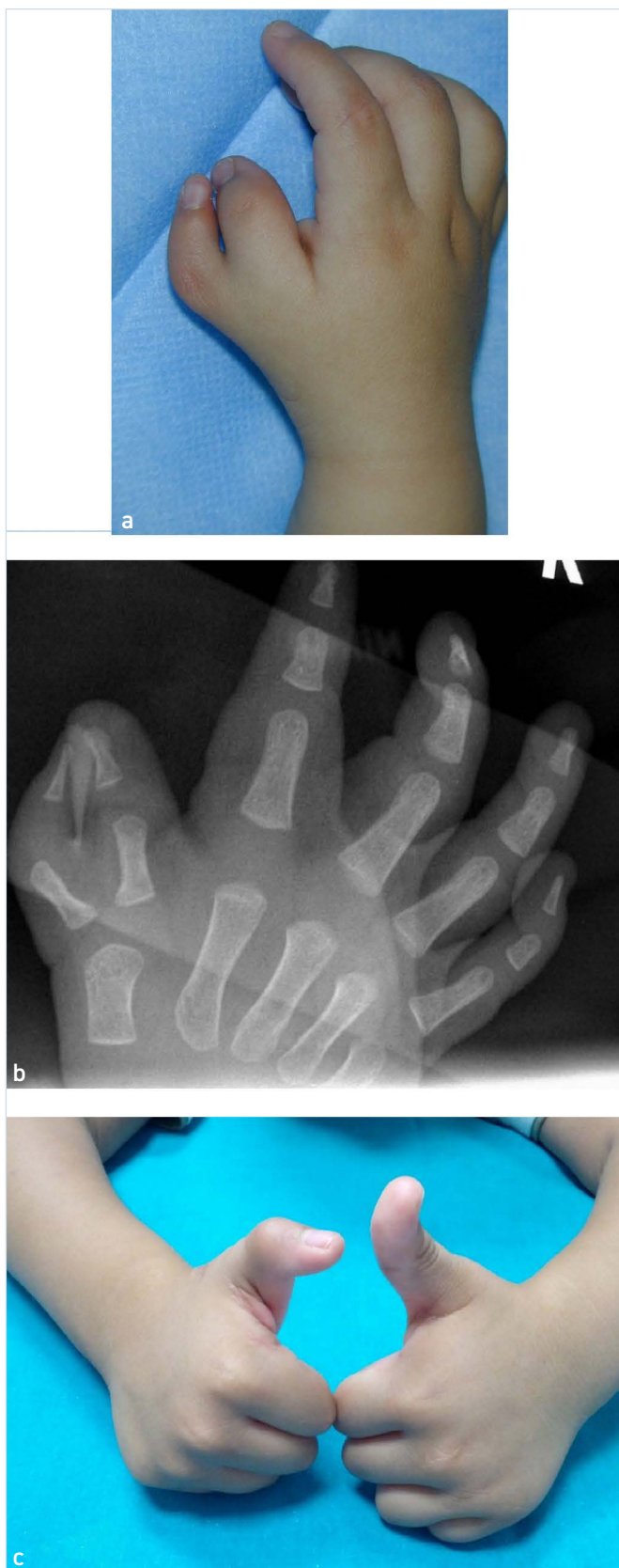


Figure 3. Patient with type IV deformity (Wassel classification), (a-b) preoperative image, operated at 15 months with removal of the supernumerary radial thumb. Clinical and radiological evaluation at 20 months post-op, showing EPL lag, but good axis realignment (c-d). Clinical evaluation after EPL retensioning and Z plasty of the first web (e-f).

Functional results in radial polydactyly have been usually evaluated using the Tada score, which is a validated functional scale that takes into account the most common complications. Apart from the Koichi scale used by Abid et al., the Tada score is used in all the studies on thumb duplication ²⁹. Although its use is usually recommended to facilitate the comparison between studies, recently Dijkman et al. showed that the JSSH assessment system provides the most reliable eva-

luation of the final results for radial polydactyly³⁰. This system includes 13 items overall: seven functional, four cosmetic and two subjective. This evaluation system will be increasingly used in future studies, possibly replacing the Tada score³⁰.

The incidence of postoperative complications (Fig. 1) is reported to be as high as 27-49%. Although most complications are minor, a second procedure is necessary in 11.5-37% of cases. Naasan and Page reported an incidence of complications of 49% in 43 patients, with the need of reoperation in 16.3% of cases. Ogino et al. reported an incidence of complications of 34% in 113 hands, with reoperation in 11.5% of cases³¹. Ozalp et al. reported an incidence of reoperation of 37.3% in 67 patients (with ligamentous reconstruction, tendon retensioning, corrective osteotomy, and interphalangeal arthrodesis)³². Cabrera González et al. treated 99 patients with the removal of the supernumerary thumb, reporting an incidence of complications of 27% and reoperation in 12% of cases¹². The results reported in this study (minor complications 19% and reintervention 14%) are similar to the literature. Patients treated for scar hypertrophy, clinodactyly and Z deformity obtained both functional and cosmetic improvement, suggesting that a corrective procedure is highly recommended for these cases.

Perez-Lopez et al. observed that the most common complications are late axial deformity, simultaneous clinodactyly and instability, with secondary procedures including collateral ligament reconstruction, tendon reattachment, and arthrodesis of the IP joint. They reported that the lowest rate of complications occurred in patients treated before 6 months of age (0% complications) or between 7 and 12 months of age (19% complications), suggesting surgery for patients aged between 7 and 12 months. Moreover, Wassel types III and IV resulted in a higher rate of complications.

Stutz et al. reported on the long-term outcomes of 41 patients with preaxial polydactyly and observed a 19% revision rate at 8 years postoperatively (mean value). The primary cause for the revision surgery was instability, which was frequently associated with pain. Revision surgeries included five IP joint arthrodesis, four tendon realignment procedures, one carpometacarpal collateral ligament reconstruction, and one opposition transfer. Larsen and Nicolai reexamined 19 patients at 22.5 years (mean value) from surgery. Seven patients were dissatisfied with the cosmesis of their thumb because of the residual angular deformity, and 14 thumbs showed > 20° of malalignment³³. Goldfarb et al. found that the unsatisfactory cosmetic appearance of the thumb after preaxial polydactyly correction was associated with residual angulation as well as a reduced nail width³⁴⁻³⁶.

The treatment of the EPL tendon lag remains challenging. All the 3 cases treated with tendon retensioning, arthrorisis with Kirschner wire and static splint experienced a provisional improvement and a deficit relapse 6 months after the

procedure (Figs. 2, 3). This complication is rarely reported in the literature. Kawabata et al. described an extensor indicis tendon transfer to the EPL tendon, performed in 6 patients aged 4-12 years, and nail dystrophy only in Wassel type I and II³⁷. The Authors consider the proper reconstruction of the lateral side of the nail mandatory and the Bilhaut-Clocquet modified technique should be preferred, in order to preserve a complete nail. The surgical treatment of congenital thumb duplications needs a solid knowledge of the anatomical variants of the skeletal segments and soft tissues, also considering the possible structural and functional changes of the thumb. The removal of the supernumerary thumb alone is rarely satisfactory. It is nearly always necessary to perform additional procedures both to correct thumb axis and stability and to obtain the best cosmetic outcome. Despite these surgical measures, the Authors' experience shows that reoperation is frequent after the removal of the supernumerary thumb (32%), as described in the literature. This proves the complexity of the malformation and the evolution of the results obtained during the developmental age. The Authors believe that the appropriate timing and the proper technique during the first procedure could lower, but not erase, the incidence of postoperative deformities and imperfections of the residual dominant thumb.

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