Type IV-D thumb duplication: A new reconstruction method

A. Abid∗, F. Accadbled, G. Knorr, P. Darodes, J.-P. Cahuzac, J. Sales de Gauzy

Pediatric Orthopaedics Unit, Children Hospital, 330, avenue de Grande-Bretagne, 31059 Toulouse cedex 9, France

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KEYWORDS
Thumb duplication; Thumb; Bilhaut-Cloquet; Wassel

Summary
Introduction: In the Wassel type IV classification category, the thumb is duplicated from the metacarpophalangeal joint; this abnormality accounts for approximately 50% of the cases of thumb duplication. Type IV is divided into four subtypes in which the IV-D type, or convergent (9% of cases), is the most complex form because both thumbs are hypoplastic with a divergent metacarpophalangeal joint and a convergent interphalangeal joint. Reconstruction is prone to axis deformity and ligament laxity, whereas the Bilhaut-Cloquet technique’s main pitfall is nail dystrophy. We propose a modified Bilhaut-Cloquet procedure to avoid these complications.

Material and method: Four males (mean age: 11 months; range: 10–12 months) with IV-D thumb duplication were operated on using a modified Bilhaut-Cloquet procedure.

Surgical technique: The skin is preliminarily marked, taking into account the excision of the central skin and the more hypoplastic nail of the two (most often the radial nail). Both proximal phalanges are split longitudinally and the central halves discarded. An oblique osteotomy is performed at the base of the distal phalanx of the ulnar thumb duplicate (the less hypoplastic) and the radial wedge is excised. The same osteotomy is applied to the distal phalanx of the radial thumb duplicate, but the radial wedge is preserved. The proximal phalanx and the bases of the distal phalanx are joined by bone suture. Axis correction and ligament stability are thus achieved without nail surgery.

Results: The patients were examined with a mean 24 months of follow-up (range: 12–36 months). The result was good in all four cases according to the Horii score.

Discussion and conclusion: This procedure combines an excision of the central part of the proximal phalanx and partial excision of the base of the distal phalanx. It provides axis correction and stabilization of the interphalangeal joint while avoiding subsequent nail dystrophy because a single nail is preserved. Preliminary results are encouraging: no axis deformity, instability or nail dystrophy has been noted. Nonetheless, the long-term results need to be evaluated.

Level of evidence: IV retrospective study.

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Introduction

Congenital thumb duplication is the most frequent abnormality of the first digit [1]. The Wassel classification delineates seven types [2], with type IV corresponding to duplication at the metacarpophalangeal (MCP) joint and accounting for more than half the cases [2]. In 1996, Hung et al. [3] proposed subdividing type IV abnormalities into four subtypes (Table 1), of which type IV-D, or convergent (9%), is the most complex form [3]. Its clinical presentation is characteristic: the two thumbs are hypoplastic and symmetrical, with divergence at the MCP and convergence at the interphalangeal (IP) joint.

Two surgical techniques are available for type IV reconstruction: the technique reconstructing one of the two thumbs, most often the ulnar thumb [4], and the Bilhaut-Cloquet procedure [5]. The technique reconstructing one of the two thumbs requires associating several surgical acts, usually with a double osteotomy in type IV-D cases [3]. The results are good except in type IV-D in which residual deformities often persist (thumb with a Z-shaped deformity) and instability [3]. The Bilhaut-Cloquet procedure, classically indicated in distal duplications, was used by some authors in proximal duplications, in particular IV-D, with good results [6–8]. However, this procedure exposes the patient to the risk of nail deformities, which can be cosmetically unsightly [9–11]. To prevent the problem of nail dystrophy, we propose a modification of the Bilhaut-Cloquet procedure and report the preliminary results of four cases.

Material and methods

Between 2006 and 2008, four boys presenting type IV-D thumb duplication were operated in the Pediatric Orthopaedics Department. There were three duplications of the left thumb and one duplication of the right thumb. No similar cases had been noted in each child’s family. No associated anomaly was found. In all four cases, the two components of the duplication were hypoplastic and symmetrical, with divergence at the MCP and convergence at the IP. In two cases, the IPs of the two components presented 30° fixed flexion deformity. Three cases had partial cutaneous fusion between the first two phalanges and in one case cutaneous fusion was complete. In all the cases, a radiographic workup of both thumbs was done. The mean age at surgery was 11 months (range: 10–12 months). The four children operated with the new technique were followed-up for a mean 24 months (range: 12–36 months). The revision protocol included a clinical and radiographic comparative analysis. The clinical results were assessed using the score reported by Tada et al. [9] and modified by Horii et al. [12]. The result was considered good if the score was greater than or equal to 5 points, intermediate if the score was between 3 and 4 points, and poor if the score was less than 3 points (Table 2).

Surgical technique

Under general anesthesia, a new clinical evaluation measured the size of the nails of the two thumbs and the contralateral thumb, the passive mobility of the MCP and the IPs, the axis defects, and assessed laxity at the IPs. We describe here the technique used in the case in which the radial thumb was the most hypoplastic (the most frequent situation). The intervention took place with a pneumatic tourniquet placed on the arm. Using a dermographic pen, we traced the dorsal and palmar incisions to be made with central cutaneous resection removing the entire nail of the radial thumb and delimiting an ungual soft tissue flap harvested from the radial edge (Fig. 1a). The nail of the ulnar thumb duplicate (the less hypoplastic of the two in this case) was therefore totally preserved. In our series, the radial thumbnail was always resected. Proximally, the incision extended to the MCP. The first phase consisted in meticulously raising the radial flap, which was dissected level with the periost of the radial thumb so as to preserve the radial neurovascular bundle. The extensor apparatus and the flexor pollicis longus tendon were explored looking for any insertion abnormalities. In our series, a shift of the insertion of the flexor pollicis longus tendon toward the side of the deviation was always present. At the first phalange (P1), a longitudinal osteotomy was done with resection of the central half following the Bilhaut-Cloquet procedure. At the second phalange (P2) of the ulnar thumb (the thumb to be reconstructed), we began with partial disinsertion of the extensor apparatus and the flexor pollicis longus tendon of the base of the P2s, where, on the radial side, an oblique osteotomy could then be done, removing a metaphyseal-epiphyseal triangle. At the base of P2 of the radial thumb, the same type of disinsertion and osteotomy was done on the radial side. In this case, the radial metaphyseal-epiphyseal wedge with its lateral-collateral ligament was preserved. The rest of P2 was resected en bloc with the entire nail of the radial thumb (Fig. 1b and 1c). We then proceeded to multilevel bone sutures along the bone using PDS® 4.0 suture. At this stage, we matched the proximal and distal phalanx bones after having reduced the radial wedge on the ulnar component. We terminated with reinsertion of the extensor apparatus with the flexor pollicis longus tendon with stitches at the metaphysis. Finally, it should be noted that no reconstruction of the pulley system was necessary. This new procedure realigns and automatically stabilizes the reconstructed IP without having to intervene on the nail, thus preventing any risk of nail dystrophy. Skin closure of the radial flap was performed with separate stitches using Vicryl rapide® 5.0 (Fig. 1d). Immobilization with a short arm cast protected the reconstructed thumb for a duration of 4 weeks.

Results

Preoperative MCP range of movement was complete in extension with 30° mean flexion (range: 25°–40°). In two cases, the IPs presented 30° fixed flexion deformity and in

<table>
<thead>
<tr>
<th>Type</th>
<th>Description</th>
<th>Frequency (%)</th>
</tr>
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<tbody>
<tr>
<td>IV-A</td>
<td>Hypoplastic</td>
<td>12</td>
</tr>
<tr>
<td>IV-B</td>
<td>Ulnar deviated</td>
<td>64</td>
</tr>
<tr>
<td>IV-C</td>
<td>Divergent</td>
<td>15</td>
</tr>
<tr>
<td>IV-D</td>
<td>Convergent (complex)</td>
<td>9</td>
</tr>
</tbody>
</table>

Table 1 Type IV subtypes according to Hung et al. [3].
Table 2  Tada score [9] modified by Horii et al. [12].

<table>
<thead>
<tr>
<th>Score</th>
<th>2 points</th>
<th>1 point</th>
<th>0 point</th>
</tr>
</thead>
<tbody>
<tr>
<td>Motion</td>
<td>&gt; 70% of the opposite side</td>
<td>50–70% of the opposite side</td>
<td>&lt; 50% of the opposite side</td>
</tr>
<tr>
<td>Laxity</td>
<td>No laxity</td>
<td>Presence of laxity</td>
<td></td>
</tr>
<tr>
<td>Thumb axis</td>
<td>&lt; 10°</td>
<td>10°–20°</td>
<td>&gt; 20°</td>
</tr>
<tr>
<td>Cosmetic</td>
<td>Acceptable</td>
<td>Moderate deformity</td>
<td>Substantial deformity</td>
</tr>
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The other two cases, flexion was 20° and 25° with complete extension. The angular deviations were a mean 42° (range: 35°–45°) at the MCPs and a mean 57° (range: 45°–75°) at the IPs. In all the cases, there was substantial laxity at the IP of the ulnar thumb. No postoperative complications were noted.

At the longest follow-up (Fig. 1e, f, g, and h), the mean MCP range of motion was 25° in flexion (range: 20°–35°) and at the IP 10° in flexion (range: 5°–20°). In all the cases, range of motion in extension was normal in the MCPs. The two cases presenting fixed flexion deformity of the IP were partially corrected, with 10° residual fixed flexion deformity in one case and 15° in the other. No axial deviation was noted. The MCP and IP joints were deemed stable in all cases. From the radiographic point of view, no growth impairment or axis defect was observed. All the parents were satisfied with the cosmetic and functional results. According to the Tada score modified by Horii [9,12], the results were judged to be good in all four cases.

Discussion

Thumb duplication is the most frequent thumb abnormality of the first digit [1,9], type IV in more than 50% of cases [1–3,11]. Few homogenous series have investigated type IV thumb duplication [6,12,13] and even fewer type IV-D [3,14]. Ulnar thumb reconstruction after excision of the radial thumb duplicate remains the most frequently used treatment today in type IV duplications, providing good results [1,9]. The poor results when using this technique are for the most part observed in type IV-D duplications in which, despite the use of a double osteotomy, there are frequent sequela such as axis deviations (thumb with a Z-shaped deformity) or instability [8,13]. Type IV-D is one of the most complex forms and the most difficult to treat [3]. The two thumbs are hypoplastic and often symmetrical, with divergence at the MCP and convergence at the IP. The IPs are unstable, often with rotational deformity and fixed flexion deformity [3]. To improve stability and the axis deviation, some authors [6–8] have used the Bilhaut-Cloquet technique in convergent type IV thumb duplications, a method usually indicated in distal type I and II duplications. This has made it possible to correct the axis defects and to stabilize the thumb, but at the cost of joint stiffness and most particularly nail dystrophy that is often unesthetic [7,9–11]. In a recent series of four cases of type IV-D duplication, Tien et al. [14] proposed a reconstruction technique using only the soft tissues and recentering the tendons, reconstruction...
of the pulley systems, and ligament reconstruction. With 3.3 years of follow-up, they have noted a 10° residual radial deviation at the IP level in one case and they have obtained a mean gain of 5° in flexion at the MCP and a mean loss of 5° at the IP. The results were deemed good in three cases and fair in one case.

In our department, we used the Bilhaut-Cloquet method in type IV-D thumb duplications with good functional results, but parents were not satisfied because of ungual dystrophy. This led us to modify the Bilhaut-Cloquet technique on the second phalange.

In our technique, we performed a central resection at the P1s with bone suture, as in the Bilhaut-Cloquet procedure. This automatically corrected the proximal divergence and stabilized the MCP at the same time. The P1 bone suture should restore the congruence of the joint surfaces of the P1 base and head, and put the proximal phalanx growth plates at the same level. Any discrepancy at this level exposes the patient to a risk of joint stiffness and growth impairment. When the proximal phalanges differ in length, a shortening osteotomy of the longest phalanx may be necessary to position the joint surfaces and growth plates at the same level. We never encountered this situation in our series. We preserved the entire nail of the less hypoplastic thumb (generally the ulnar thumb duplicate) and we resected the metaphyseal-epiphyseal triangular wedge at the base of P2. At P2 of the thumb to be excised, only the lateral metaphyseal-epiphyseal wedge, the collateral ligament, and the soft-tissue flap were used to construct the second phalange of the reconstructed thumb. Thus, we obtained realignment and automatic stabilization of the IP without risking nail dystrophy. However, the width of the less hypoplastic nail should be greater than or equal to 70% of the width of the contralateral thumbnail for a good cosmetic result. We believe this is a limitation in this technique. In our series, the MCP retained good flexion with a mean loss of 5°. Stiffness was greater on the IP joint, but this did not or only slightly disturbed thumb function. Moreover, a stiff but well-aligned IP is more functional than a flexible but deviated and unstable IP. In our experience, the growth disturbance described with the Bilhaut-Cloquet technique is very rare and can be, in our opinion, prevented with more rigorous technique.

Conclusion

In the simple forms of type IV thumb duplication, reconstruction of one of the two thumb duplicates (most often the ulnar thumb) remains the choice treatment in our view. However, in complex type IV-D thumb duplications, this technique is insufficient because often an axis deviation and instability persist. In these forms, we prefer to use the modified Bilhaut-Cloquet procedure whose preliminary results are encouraging, notably because of the absence of residual nail dystrophy. This needs to be confirmed by a longer-term study.

Conflict of interest statement

None.

References