On-Top-Plasty for Atypical Thumb Duplication: A Case Report With 10 Year Follow-up

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Abstract

Background: A very uncommon pattern of thumb duplication consists of an ulnar-side floating thumb and a radial-side distally hypoplastic thumb. **Methods:** We report the case of a 15-month-old child with this type of thumb duplication on his right hand treated with an on-top-plasty technique. The ulnar-side segment was pedicled and transferred to the lateral thumb, which was distally resected. **Results:** A well-aligned and widely mobile thumb with a wide first web and an excellent cosmetic appearance was exhibited 10 years after surgery. **Conclusions:** On-top-plasty technique might be amenable to reconstruct certain atypical thumb duplications.

Keywords: thumb duplication, preaxial polydactyly, congenital hand, on-top-plasty

Introduction

Thumb duplication is one of the most common congenital upper limb anomalies.^{1–3} A few authors have described a very uncommon pattern of thumb duplication, not classifiable by Flatt's modified Wassel's classification system, and accounting for a mere 0.7% of thumb duplications.^{1,3–6} It consists of an ulnar-side floating thumb and a radial-side distally hypoplastic thumb^{1,4,5} (Figure 1).

Methods

We report the case of a 15-month-old child with this type of thumb duplication on his right hand. The radial thumb was distally hypoplastic and deviated, but had a well-developed and mobile basal joint (Figure 1); meanwhile, a cosmetically acceptable ulnar thumb floated in the first web, exhibiting a proximally absent metacarpal (Figure 1).

Following the treatment principle of “spare parts surgery,” we selected to perform an on-top-plasty of the ulnar thumb over the radial thumb, after resecting the latter's hypoplastic distal segment.¹

A curved palmar incision, including a racquet incision for both thumbs, was performed. First, dorsal veins and volar neurovascular bundles were identified for the ulnar thumb, allowing it to be pedicled for subsequent lateral transfer (Figure 2a). Second, resection of the distal lateral thumb was performed at the level of the distal metaphysis of its metacarpal (Figure 2a). Finally, a pedicled on-top-plasty of the ulnar thumb over the residual lateral thumb was performed, and pinned with a 1-mm

Kirschner wire (K-wire) (Figure 2b). The adductor pollicis muscle was inserted into the ulnar thumb. Abductor pollicis brevis was inserted in the lateral thumb, hence requiring detachment and reattachment into the ulnar thumb. Flexor pollicis longus tendon was absent in both thumbs. Extensor tendons, present in both thumbs, were sutured to each other after the on-top-plasty. Postoperatively, a thumb spica cast was placed, which was removed, together with the K-wire, 4 weeks after surgery. No formal therapy was instated afterwards.

Results

Ten years after surgery, the patient exhibited a well-aligned and widely mobile thumb with a wide first web and an excellent cosmetic appearance (Figure 3, Video 1). The patient also demonstrated both gross and fine thumb motion, with the affected (right) hand dominant. Skeletal length was 71 mm in the involved thumb versus 80 mm in the contralateral thumb (Figure 3), a difference that was almost unnoticeable. Thumb pinch strength was 3 kg for the involved thumb and 4

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Figure 1. Atypical thumb duplication showing a distally hypoplastic and deviated radial thumb (a) but a well-developed basal joint (b). Conversely, the ulnar thumb exhibits a proximally absent metacarpal (b) but a well-developed distal thumb that looks normal (a, b).

Figure 2. The ulnar thumb is pedicled on the neurovascular bundles while the distal radial thumb is resected (a). Then, the ulnar thumb is transposed to the residual radial thumb and fixed with a Kirschner-wire (b).

kg contralaterally. The parents and the child were very satisfied with both the functional and cosmetic results.

Discussion

Three strategies have been described for the treatment of this uncommon thumb duplication. Ogino et al described

a simple translocation technique that consists of transferring the ulnar thumb to the radial thumb using a lateral approach and a racquet incision, thereby obviating any need to dissect the ulnar thumb vessels.⁴ Later, this technique was used by Shen et al in 5 consecutive patients, yielding excellent short-term functional and cosmetic results.⁵ This procedure was deemed inappropriate for our

Figure 3. Follow-up 10 years after surgery. Radiography reveals adequate longitudinal growth of the reconstructed thumb (a), and the patient had a satisfactory functional and cosmetic result (b).

patient, however, because it would have resulted in a very narrow first web space, as the ulnar thumb was very close to the index finger.

Dobyns et al reported a pedicled “on-top-plasty” for the management of this type of thumb duplication but not specific postoperative information regarding functional and cosmetic results was provided.¹

Finally, Shen et al recommended excising the ulnar thumb if no active motion was observed, preserving the radial thumb rather than using the translocation technique.⁵

Conclusions

The on-top-plasty technique might be a satisfactory procedure for treating this particular pattern of thumb duplication. However, this technically demanding procedure might be not necessary if a wide space is already present between the ulnar thumb and the index finger, in which case the translocation technique might be preferable. A third option might be resecting the floating ulnar thumb and preserving the radial thumb. However, though this is the simplest solution technically for this deformity, it also yields the least cosmetically pleasing result and may cause functional difficulties.

Ethical Approval

In advance of any data collection, the protocol of the present study was approved by the local ethics committee.

Statement of Human and Animal Rights

All procedures in this study were in accordance with the Declaration of Helsinki guiding biomedical research involving human subjects.

Statement of Informed Consent

Written informed consent before their participation was obtained.

Declaration of Conflicting Interests

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