

Reversed vascularized second metatarsal flap for reconstruction of Manske type IIIB and IV thumb hypoplasia with reduced donor site morbidity

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Abstract

Background: The predominant method for Manske type IIIB and IV thumb hypoplasia is pollicization. However, for those who are not willing to sacrifice the index finger, a method that could reconstruct a functionally capable and aesthetically acceptable thumb remains desirable. This study aimed to investigate and assess the functional and radiographic outcomes of utilizing a reversed vascularized second metatarsal composite flap for thumb reconstruction as a new alternative.

Methods: From May 2014 to January 2017, 15 patients with Manske type IIIB or IV thumb hypoplasia who were admitted to the Department of Hand Surgery, Beijing Jishuitan Hospital were included in this study. An osteocutaneous flap containing a section of second metatarsal and its distal head was transferred in reversed position to reconstruct carpometacarpal joint. The donor site was reconstructed by a split half of the third metatarsal. Various functional reconstructions were commenced at second stage. The reconstructed thumbs were evaluated using the Kapandji score, pinch force, and the capacities of performing daily activities through a detailed questionnaire.

Results: Among these 15 patients (seven type IIIB and eight type IV), there were ten boys and five girls with median age of 4.2 years (range: 2.0–7.0 years). There were seven right, three left, and five bilateral thumbs for whom only the right thumb received surgery. There were 14 metatarsal flaps survived (14/15). With an average follow-up of 19.2 months, the reconstructed thumbs had acceptable functional and aesthetic outcomes and the donor foot presented in decent appearance without signs of impaired function. All 15 children have improved the Kapandji score (from 0 to an average of 6.7), pinch force (from 0 to an average of 1.5 kg), with ability of grip and pen holding. X-ray indicated continuous bone growth. Patients and parents had good acceptance of the new thumb.

Conclusions: Reconstruction of an unstable hypoplastic thumb (Manske type IIIB and IV) with use of a vascularized metatarsal is an effective strategy. It offers an alternative solution for parents insisting on saving the thumb.

Keywords: Thumb hypoplasia; Second metatarsal flap; Donor site morbidity; Microsurgery

Introduction

Manske type IIIB and IV thumb hypoplasia requires complex reconstructive procedures because they represent a major disability.^[1] To date, the most established surgical intervention is ablation and index pollicization,^[2,3] as it has been widely perceived that the results of other methods may not be as good as pollicization.^[4-6] However, finger ablation is not always accepted by parents. In our opinion, the impact of a mutilated hand that might negatively affect those children's emotional and psychological development cannot be neglected. Ideally, a method that could reconstruct the hypoplastic thumb, without major donor

site morbidities, and at the same time, obtain acceptable aesthetic outcome, remains still desirable.

This study brings forth a new surgical design that could overcome some shortcomings of the previous reconstructive methods, assessing its functional, radiographic, and patient-perceived outcomes, and attempt to offer a new solution for the patients and parents. The core of this surgical modality is to utilize a vascularized second metatarsal osteocutaneous flap to reconstruct the skeletal framework of the first metacarpal and carpometacarpal joint, as well as to provide additional skin envelope to the underdeveloped thenar area. Secondary procedures that aim to improve the thumbs functions will commence

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during the staged surgeries. In addition, a split of the adjacent third metatarsal with vascularity is transposed to reconstruct the donor site.

Methods

Ethical approval

The study was conducted in accordance with the *Declaration of Helsinki* and was approved by the Institutional Review Board of Beijing Jishuitan Hospital. Surgical plans were illustrated in detail to the parents, potential complications and shortcomings, including the possibility of a functionally compromised thumb were also discussed. Informed written consent was obtained from all parents before their children enrollment in the study.

Patients information

From May 2014 to January 2017, 15 patients who were admitted to the Department of Hand Surgery, Beijing Jishuitan Hospital were included in this study. All patients had been diagnosed with Manske type IIIB or IV thumb hypoplasia, and their parents' insisted on preserving all five fingers. Physical examinations suggested that all patients had some degree of radial inclinations at the interphalangeal (IP) joints, and the metacarpophalangeal joints were severely underdeveloped. The patients who were less than 2 years old or with severe comorbidities were excluded.

Surgical technique

Hand preparation

A longitudinal incision, 4 to 5 cm in length, was made radially close to index metacarpal and located between the radial styloid proximally and the radial aspect of the dorsum of the floating thumb distally. The underdeveloped first metacarpal was trimmed to normal bone and index metacarpal base was exposed. The underdeveloped muscles and ligaments around the carpometacarpal joint were released to make adequate space for placement of the flap. Pollex abductus of the hypoplastic thumb at radial side of the IP joint was detached at the proximal insertion to release the joint, and correct radial inclination of the IP

joint. Seven cases had an underdeveloped trapezium, all of which had the abductor pollicis longus (APL) and extensor pollicis brevis (EPB) attached on; these two tendon insertions were released and later sutured to the newly reconstructed carpometacarpal joint. A test pull was applied to the thumb to estimate the required length of the bone defect [Figure 1A and 1B]. In cases when the cephalic vein was absent, one major dorsal vein would be the alternative. The optimal recipient artery would be the terminal branch of radial artery at the thumb base; when it was unavailable, the common digital artery between the index and middle finger or superficial palmar arch was prepared with sufficient length then transposed for anastomosis.

Second metatarsal flap harvest and donor site reconstruction

An elliptically-shaped flap was designed at the dorsum of second metatarsal. The flap was as wide as possible while still allowed direct closure. The length of the flap usually slightly exceeded the second metatarsophalangeal joint distally and second tarsometatarsal joint proximally. An "S"-shaped incision was made to explore the dorsal vessels [Figure 2A]. Medially, the great saphenous vein and its branches into the flap were dissected, while the lateral and distal venous branches were ligated. For artery preparation, medial to the first metatarsal, both the distal proper digital artery and proximal dorsalis pedis artery were prepared. The branch derived from dorsal metatarsal artery supplying second metatarsal head, all lateral branches supplying the skin, and importantly, the arcuate artery were preserved. The joint capsule and ligaments were divided and an osteotomy was performed, rendering an osteocutaneous flap [Figure 2B and 2C]. The length of bone was based on the measurement from previous step.

For reconstruction of the donor site, since the third metatarsal maintained its blood supply from the arcuate branch, we used one of the following two approaches. The third metatarsal was exposed, split longitudinally, and transposed to the second metatarsal defect. A simpler method was to incompletely split the third metatarsus into a "V" shape and interpose the medial half into the second metatarsus defect [Figure 2D]. A Kirschner wire (0.8 mm)

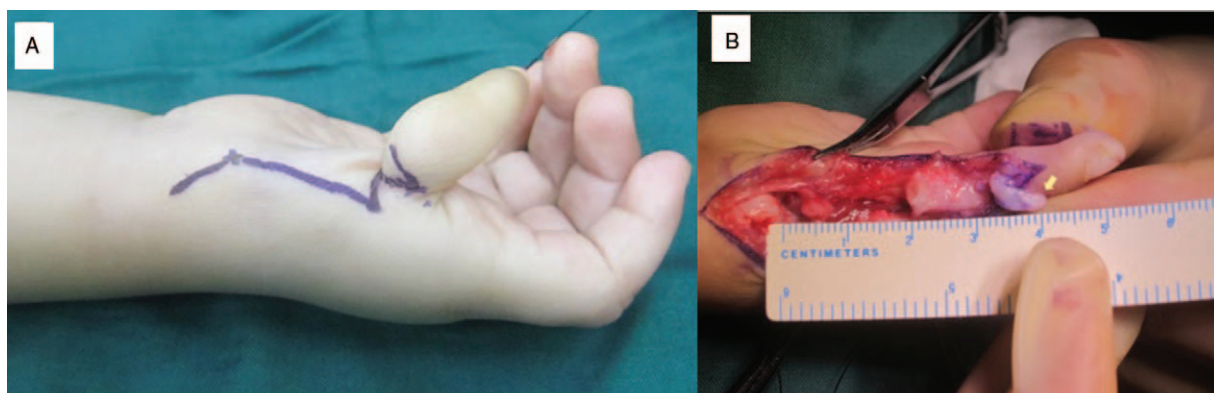


Figure 1: Hand preparation: (A) incision design; and (B) the dissection and measurement of the deficiency (arrow showing the pollex abductus).

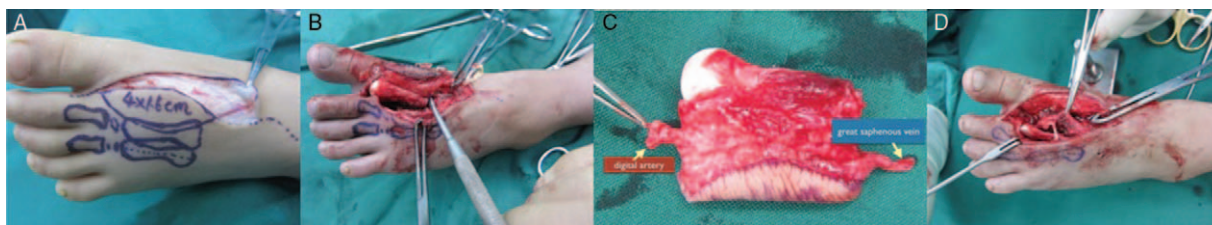


Figure 2: Flap harvest and donor site closure: (A) flap design and dorsal incision; (B and C) the second metatarsus is dissected and osteomized, rendering an osteomyocutaneous flap; (D) the third metatarsus is split and transposed to reconstruct the resultant defect.

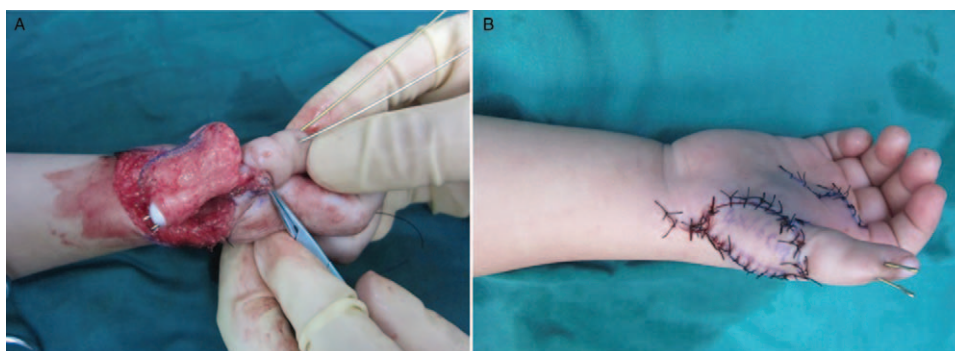


Figure 3: Thumb reconstruction: (A) the metatarsus is reversely placed, anastomosed, and fixated for reconstruction; (B) wound closure.

was used to stabilize the bone stumps, together with absorbable sutures (3-0 PDS-II) to further consolidate the surrounding soft tissue. The wound and skin were closed primarily. A plaster cast was applied to hold the foot in a functional position, and was later replaced by a customized foot brace.

Thumb reconstruction

The thumb was stretched into abduction and pronation. For those patients with an extremely limited thumb web space, the skin envelope from the flap was still insufficient. In such cases when full thumb abduction was not possible, we would gradually adjust the thumbs to match skin envelop. Two intra-medullary Kirschner wires were channeled from the first metacarpal remnant through the thumb tip for later fixation. Next, the composite flap was reversed, so that the metatarsal head was proximally placed into the defect of the thumb base [Figure 3A]. The two previously placed Kirschner wires were advanced through the metatarsal head into the second metacarpal base or trapezium. When this fixation was not possible, the metatarsal head was firmly sutured to the trapezium or second metacarpal base. Noted that the thumb was fixated slightly in over-pronation, this was to achieve better opposition, particularly a pulp-to-pulp opposition between the reconstructed thumb and the other fingers. Then, the metatarsal joint capsule was sutured to soft tissue around trapezium cartilage [Figure 3B]. When the insertion of the APL and EPB existed, they would be re-fixated to the newly reconstructed joint.

Vessels for anastomosis were the dorsalis pedis artery to the residual radial artery (seven cases) or the common digital artery (eight cases); the great saphenous vein to

cephalic or dorsal vein using 11-0 monofilament nylon suture under the microscope [Figure 4].

The post-operative protocol included anti-coagulants (heparin saline, 5000 U/100 mL), prophylactic antibiotics (cefuroxime sodium, 100 mg/kg) for 3 days, and fluid support. Any sign of insufficient blood flow to the flap was an indication for immediate revision surgery. A wrist brace was mandatory for 6 weeks, and foot brace was used for 8 weeks. Parents were asked to regularly abduct the reconstructed thumb as rehabilitation by the time the Kirschner wire was removed until the secondary surgery.

Second stage reconstruction

At minimal of 4 months post-operatively, secondary reconstructions were conducted. The procedures were tailored to each case including flexor digitorum superficialis (ring finger) transfer to improve thumb abduction; thumb-index web space release by local flap transposition and Z-plasty; fat harvested by liposuction was transferred to enhance thenar area fullness. Rehabilitations were commenced afterward.

Post-operative assessment

The evaluations were radiographs of hand and donor foot at follow-ups, daily activity performance evaluation including the ability to grab small and large objects, unintentional incorporation of thumb, and general impression on the overall treatment as previously described.^[7] The detailed visual analog scale (VAS) questionnaire was completed by parents at 6 months of follow-up [Supplement material, <http://links.lww.com/CM9/A102>].^[8]

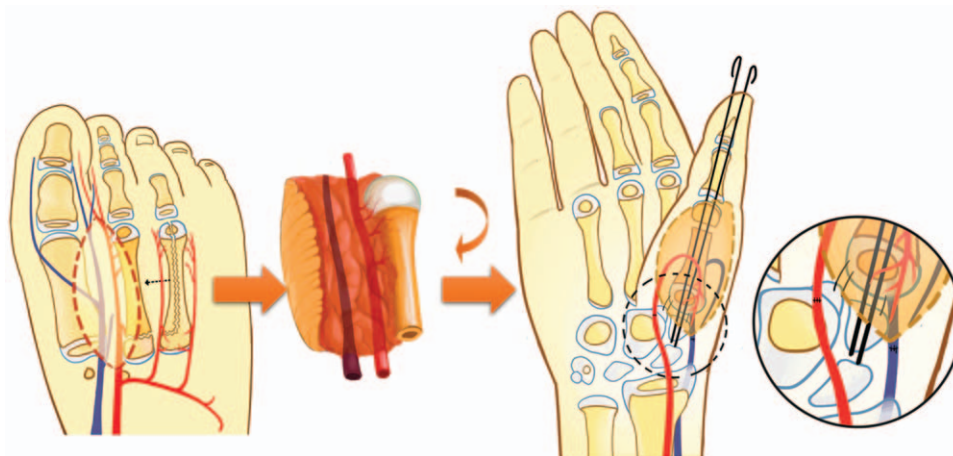


Figure 4: Surgical schematic of utilizing reversed vascularized second metatarsal flap for reconstructing hypoplastic thumb.

Results

Among these 15 patients (seven type IIIB and eight type IV), there were ten boys and five girls with median age of 4.2 years (range: 2.0–7.0 years). There were seven right, three left, and five bilateral thumbs for whom only the right thumb received surgery. In one patient, the surgeon at the local hospital excised one of the duplicated thumbs before referring to us (case No. 2); one case had Holt-Oram syndrome with a ventricular septal defect, club hand on the right side and microtia (case No. 5); one case had radial deficiency (case No.10) [Table 1]. The flap size ranged from 1.5 cm × 3.0 cm to 2.2 cm × 5.8 cm (average: 1.8 cm × 4.5 cm). There were 14 (14/15) metatarsal flaps survived. Two patients had minor skin necrosis on the edge at donor site due to excessive tension, healed by non-operative measures. For the 14 flaps, post-operative radiographs at 2-month indicated bone union and growth [Figure 5].

However, for one case (1/15), the flap had compromised blood supply shortly after the operation and eventually turned into skin necrosis with delayed bone union at the reconstructed metacarpus. This patient received a pedicled vascularized flap on the dorsal-radial side of the wrist and iliac crest bone grafting. No other morbidities were noted afterward.

At an average follow-up of 19.2 months (range: 12–34 months), the reconstructed thumbs and donor foot had close to normal appearance, with clear epiphysis line, and continuous bone growth and thickening [Figure 6]. All 15 children have improved the Kapandji score (from 0 to an average of 6.7), pinch force (from 0 to an average of 1.5 kg), with ability of grip and pen holding. Still, three patients had difficulties with delicate movements, two children had inadequate Pulp-to-Pulp pinch. The donor foot presented in good appearance without any signs of impaired function, X-ray further evidenced good bone fusion in second and third metatarsus [Figure 6]. Patients' daily physical activities like walking, running, and jumping were not affected. Detailed VAS results were summarized in Supplemental material, <http://links.lww.com/CM9/A102>. Briefly, although with some degree of functional imper-

fections, parents had acceptable recognitions on the new thumb, with inclinations to recommend this treatment to others.

Discussion

Thumb hypoplasia refers to a spectrum of clinical abnormalities ranging from a slightly small digit to complete absence of the thumb unit.^[9] The goal of treating such abnormality is to provide the child with a stable and functional thumb unit, as well as to improve cosmesis.^[10] In spite of multiple studies have suggested better functional outcome of pollicization than what various bone flap based reconstructions could achieve,^[11,12] parents' objections to amputation are strong in China, as finger mutilation is associated with criminality in the afterlife by the social perceptions. Speaking from our experiences, most parents would not hesitate to trade certain degree of functional inadequacy for a whole-looking hand.

The determinant features of Manske type IIIB and IV hypoplastic thumb are proximal metacarpal absence and instability at the carpometacarpal joint,^[13] which also lay out the key challenges for reconstructions. Our solution is to first reconstruct the skeletal framework of carpometacarpal joint by utilizing the distal half of the second metatarsal. This flap has two advantages: (1) the skin flap signifies the blood supply, as an indicator for the success of the transplantation after the operation; (2) the absent thenar muscles and underdeveloped thumb metacarpus is associated with insufficient skin envelope, a composite flap could provide additional area of skin for primary wound closure as well as to facilitate the second-stage procedures.

The advent of supermicrosurgery brings new breakthroughs on the technical level, in addition, several studies have contributed rationalizing our procedure: first, post-transplant epiphysis still maintains the growth potential.^[14-16] It has been confirmed that vascularized epiphysis had no histological alterations when compared to the original.^[17] This offered some insights on how the vascularized second metatarsal still retained its potential that eventually turned into a continuously growing

Table 1: Information of all patients with Manske type IIIB and IV thumb hypoplasia in this study.

Case No.	Gender	Age (years)	Type/side	Flap size (cm × cm)	Complications	Staged reconstruction	Kapandji Score	Pinch force (kg)
1	Male	7.0	IIIB/left	1.8 × 4.0	N/A	MLR, FT [*] , thenar plasty	6	1.00
2	Male	2.0	IIIB/right; IIIA/left	1.5 × 3.0 (right)	N/A	MLR, FT [*]	4 (right)	0.50 (right)
3	Female	5.0	IIIB/right	1.5 × 3.0	Minor foot skin necrosis	MLR	10	5.00
4	Male	5.0	IV/right; IV/left	2.0 × 4.0 (right)	N/A	MLR, thenar plasty [†]	9 (right)	4.00 (right)
5	Female	3.0	IV/right; IV/left	1.5 × 5.5 (right)	N/A	MLR	6 (right)	1.00 (right)
6	Male	3.0	IV/right; IIIA/left	1.6 × 4.0 (right)	N/A	MLR	4 (right)	0.50 (right)
7	Male	3.0	IIIB/left	1.8 × 5.5	N/A	MLR, FT [*] , thenar plasty [†]	9	1.50
8	Male	4.0	IIIB/right	1.6 × 5.0	N/A	MLR, osteotomy, FT [*] , thenar plasty [†]	10	3.00
9	Female	3.0	IV/right	2.0 × 4.5	N/A	MLR	3	0.50
10	Female	5.0	IV/right	2.2 × 5.8	N/A	MLR	8	0.75
11	Female	4.0	IV/right	2.0 × 5.0	Skin necrosis, delayed bone union	Pedicled flap transfer, bone graft fusion, MLR, thenar plasty [†]	9	1.80
12	Male	4.0	IV/right; II/left	1.8 × 4.5 (right)	N/A	MLR	6 (right)	1.00 (right)
13	Male	3.0	IV/right	1.8 × 4.5	N/A	MLR, FT [*]	5	0.75
14	Male	5.0	IIIB/left	1.9 × 5.0	Minor foot skin necrosis	MLR, FT [*] , thenar plasty [†]	5	1.00
15	Male	3.0	IIIB/right	1.8 × 4.5	N/A	MLR, thenar plasty [†]	7	0.50

*Fat that was harvested by liposuction from the abdomen and then infused to the thenar area to enhance its appearance, †a series of Z-plasty and local flap transfer to release the web space between thumb and index finger. N/A: Not applicable; MLR: Metacarpophalangeal ligament reconstruction; FT: Fat transfer.

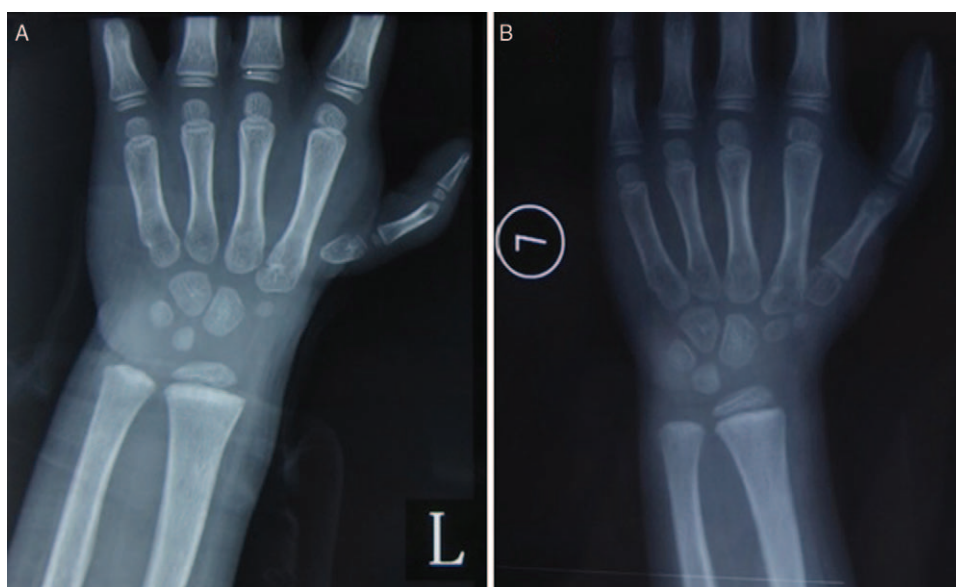


Figure 5: Pre-operative (A) and 1-year post-operative (B) X-ray of a Manske type IIIB thumb hypoplasia. The reconstructed thumb presented satisfactory growth and development.

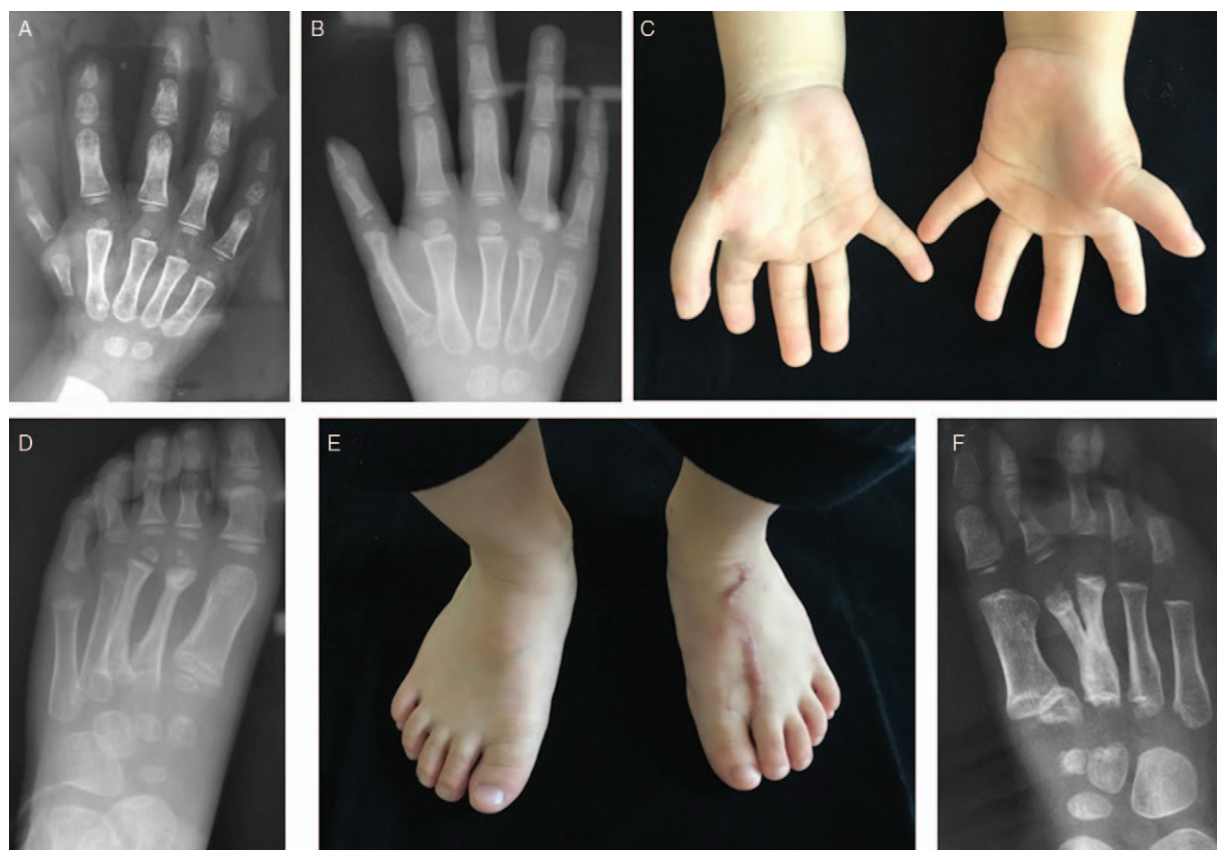


Figure 6: Post-operative evaluation: (A–C) 1-year after reconstruction, the thumb presented in good appearance, with X-ray demonstrated satisfactory metacarpal growth; (D and E) recovery of the donor foot; (F) another patient whose foot was repaired with V-split third metatarsus.

metacarpus; the same mechanism applies to the split metatarsus being self-repaired as well.

A vascular study of Petersen *et al*^[18] suggested that the metatarsal heads contains two arterial supplies: (1) the dorsal metatarsal arteries from the dorsalis pedis artery, and (2) the plantar metatarsal arteries, which are the branches of the posterior tibial artery. These two vessels anastomose at the metatarsal heads, forming a vascular ring. Such interconnections ensure sufficient blood flow to the metatarsus as long as one of the two vessels is preserved. In our series, the donor site was supplied by dorsalis pedis-dorsal metatarsal arteries; all lateral branches derived from first dorsal metatarsal artery to the second metatarsal were carefully preserved. One patient who experienced some complications due to compromised blood supply might attribute to the fact that her foot vasculature was Gilbert type III, which also explained how bone graft survived despite of necrotic skin.

Another equally important aspect is to minimize foot morbidities. Some literatures have already suggested that the donor site morbidities were being underestimated,^[19] which implied extra significance to children. The strategy of utilizing a split metatarsus has been reported to repair various defects with excellent donor site recovery.^[20] In our cases, the arcuate branch was preserved, which made the adjacent third metatarsal retaining relatively uncompromised blood supply. Then, partial osteotomy or

displacement would be a straightforward approach and as well as time-efficient. To date, no patients have presented any signs of floppy unstable toes or visible deformity with growth, as evidenced by radiographs that both second and third toes have normal development, with solid cortex and epiphysis growth owing to the good vascularity.

Continuous effort utilizing composite graft for congenital hypoplastic thumb reconstruction has been reported: Shibata *et al*^[21] introduced the method of a vascularized metatarsophalangeal joint; there are three major shortcomings regarding these techniques: (1) as a saddle joint, carpometacarpal joint has far more mobility comparing to metatarsophalangeal joint^[22]; (2) their placement of transplanted metatarsophalangeal joint was not *in situ*, but distal to the original carpometacarpal location, which further limited its functions; and (3) the management at the donor site were unstandardized, the loss of metatarsophalangeal joint would inevitably hamper the full functional recovery of the foot. Tu *et al*^[7] harvested the entire second toe-metatarsal bone, which also has yet to address the negative impact on the foot.

The main hurdle of practicing this surgery is the extremely refined technique. For one, infants' vessels are small and thin in nature, and the hypoplastic thumbs usually associate with abnormal vasculature, all abound with increased risks. Therefore, expertise in microsurgical or

super-microsurgical skills, along with extensive knowledge in anatomy is mandatory for the consistent success. Nevertheless, a backup plan is always necessary, such as switching to pollicization or other measures when reconstruction is deemed impossible intra-operatively. Furthermore, the operation usually takes considerably long time: in our department, even with two teams working simultaneously, 4 to 7 h are routinely required. In addition, staged reconstruction for muscle and tendon reconstruction is necessary, for which, parents need to be fully informed and educated with the arrangement.

Two patients' parents were somewhat unsatisfied with the results, which paralleled the poor performance scores of the reconstructed thumbs. This might be due to the high expectations they initially held. One must realize that the limitations of this method before committing to it. A reconstructed thumb that has had little to no functions from birth would almost certainly have intrinsic disadvantages when comparing to the one that is transposed from pollicization. On the other side, these parents were not fully compliant with the rehabilitation during follow-ups, which could be another factor that hampered the best outcome possible. Theoretically speaking, the somatosensory cortical plasticity comes into effect after reconstruction,^[23,24] constant stimuli, rehabilitation, and parental guidance would be of paramount importance. Nevertheless, when asked whether they would recommend this treatment to other fellow parents, these parents gave the score of 6 (out of 10), suggesting the likelihood of doing so. In the long run, such flawed result offers a reminder for both physicians and parents in weighing the risk-benefit balance between pollicization and reconstruction.

This reconstructive technique provides a new approach to reconstruct a functionally capable and cosmetically acceptable thumb that also retains growth potential, with reduced donor site morbidities. Nevertheless, more cases are required. Long term follow-up, as well as more systemized and objective evaluation measures are necessary to further demonstrate its place when comparing to pollicization. But again, the preliminary success has exemplified that this approach can be performed effectively despite the technical challenges. We are optimistic that it could become a mainstay method for hypoplastic thumb reconstruction when parents insist on "Five" rather than "Four."

Conflicts of interest

None.

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