

Can on-top plasty for atypical radial polydactyly realize a functional reconstruction corresponding to appearance?

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ABSTRACT

On-top plasty may be the best option for reconstructing atypical radial polydactyly, consisting of a developed distal portion in one thumb and a developed proximal portion in the other thumb. However, it is unclear if patients can gain thumb function corresponding to the reconstructed appearance. We performed a retrospective bibliographic review to summarize the current literature and present our department's on-top plasty experiences. Over the last ten years, our department performed three on-top plasty radial polydactyly reconstructions. We surveyed the appearance and use of the reconstructed thumb and the patient's satisfaction level during postoperative follow-up. We also retrieved published case reports describing on-top plasty for polydactyly reconstruction to compare with our cases. Our patients had an acceptable thumb appearance and were satisfied with the gained function of the thumb. However, two patients required revision surgery. We found 11 studies in the literature describing on-top plasty for polydactyly treatment, four of which reported a limited range of thumb motion, but no patient or their family complained about the result. Most patients who undergo on-top plasty for radial polydactyly can expect a fair to good appearance after surgery. Although instability or limitation of the joint arc is possible, the reconstruction satisfies patients regarding function and appearance.

Keywords: on-top plasty, radial polydactyly, function of the reconstructed thumb, patient related outcome for the child

Abbreviation:

PedsQL: Pediatric Quality of Life Inventory

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INTRODUCTION

Radial polydactyly is usually treated by ablating the lesser thumb, then reconstructing using the remaining dominant thumb. However, this method cannot treat atypical polydactyly. For example, in several unique polydactyly cases, the dominant thumb is abnormally configured, and resecting the lesser thumb is insufficient for functional reconstruction. Such cases are treated with spare

Received: November 10, 2021; accepted: February 4, 2022

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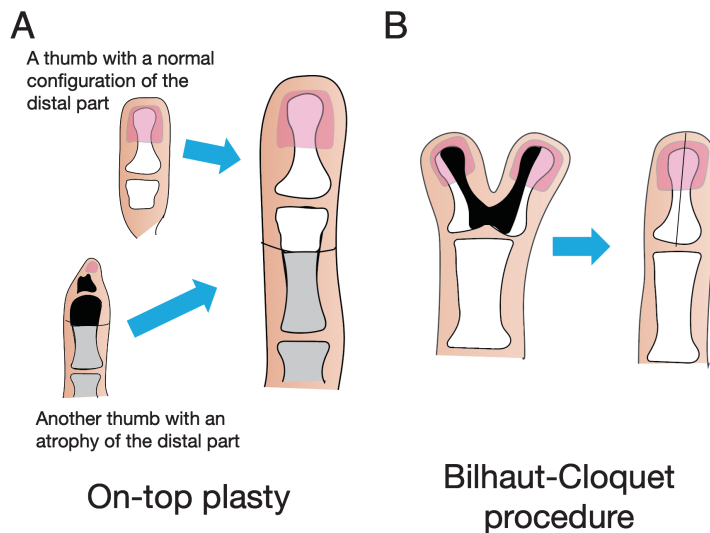


Fig. 1 Schematic figures of procedures for polydactyly reconstruction

Fig. 1A–B: Figures illustrates schematics of the on-top plasty (A) and the Bilhaut-Cloquet procedure (B).

part surgery, using both of the duplicated thumbs.

Spare part surgery for polydactyly is planned based on the morphology of both thumbs. The Bilhaut-Cloquet procedure can be used for symmetric thumb duplication, which involves reconstruction with half of each thumb (Figure 1). On-top plasty is performed when the surgeon needs to use both the proximal part of one thumb and the distal part of another thumb (Figure 1). Because of the rare incidence of such atypical polydactylies, these procedures are infrequently performed.

Over the last decade, several congenital deformity cases treated with on-top plasty have been published.¹⁻¹⁰ During this period and considering these examples, we treated consecutive cases of special duplicated thumb with spare part surgery by reconstructing with the on-top plasty method, which seemed ideal for appearance and function in these instances.

All authors publishing case series regarding thumb polydactyly reconstruction with on-top plasty agreed that the thumb appearance was good. However, it remains unclear if surgery also provides functional reconstruction. We hypothesized that reconstructing the thumb functions to patient satisfaction would be difficult. Thus, we reviewed our cases and the literature regarding appearance and function to provide clarity.

METHODS

We retrospectively reviewed thumb polydactyly reconstruction cases using on-top plasty in our hospital. The radial thumb of all patients had a normal metacarpophalangeal joint, and their ulnar thumb had a normal interphalangeal joint with nearly normal nail components. Written informed consent was obtained from the parents of the patients, who also agreed to publication. This research has been approved by the IRB of the authors' affiliated institutions.

All patients were subjectively and objectively evaluated. The first author independently asked the patients and their parents about pain, numbness, and daily life inconveniences as subjective

assessments. The active range of motion and joint instability were also evaluated. At the latest follow-up, the patients' parents were asked about the treatment satisfaction level using the face scale,¹¹ and they answered the Japanese version of the Pediatric Quality of Life Inventory (PedsQL) Generic Core Parent proxy-report,^{12,13} which is a validated approach to measure health-related quality of life in healthy children, and parents can answer by proxy.¹³

For the literature review, we retrieved scientific papers and case reports using PUBMED, EMBASE, and Ichushi-Web. Literature retrieval was performed by the first and second authors in May 2021. The search terms included "Polydactyly," "Spare part," and "On-top plasty." We excluded papers written in languages other than Japanese or English. After reading the full text, we excluded trauma cases, those other than radial polydactyly, and those who underwent the Bilhaut-Cloquet procedure. We also excluded studies reporting a variety of reconstruction procedures where on-top plasty cases could not be identified. After identifying all reported cases of on-top plasty, we evaluated the outcome of the reconstructions.

Surgical procedure

Surgery was performed under general anesthesia. Using a tourniquet, a palmar zigzag incision was made to identify the neurovascular bundle to the ulnar thumb, and it was dissected to the palmar arch. The distal part of the head of the proximal phalanx of the ulnar thumb was elevated as an island flap, including the nail (Figure 2). After elevation, the ulnar thumb was temporarily sutured to the radial side of the radial finger to confirm the tension of the neurovascular bundle after the transition of the flap. With an additional dissection of the proximal defect, the pedicle of the island flap can be extended to about 12 mm. Because an excessive prolongation of the neurovascular bundle can cause spasm of the artery and hemodynamic instability of the flap, we usually limit the extension to 5 to 7 mm. In case that a further extension is required for the placement of the flap, the shortening of the proximal phalanx is performed. The radial thumb was osteotomized at the level of the proximal phalanx to prepare the donor. After releasing the tourniquet and confirming that the blood flow to the ulnar thumb flap was maintained, the distal

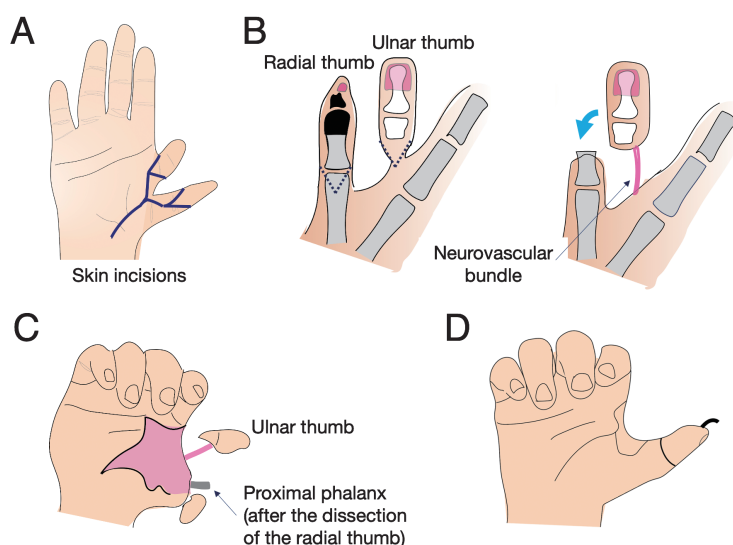


Fig. 2 The surgical procedure flow

After dissecting the neurovascular bundle, the ulnar thumb is transferred on top of the radial thumb.

part of the radial thumb was resected, and its stump was exposed. The ulnar thumb was fixed on top of the radial thumb using a Kirchner wire as an on-top plasty. The wound was closed so that the first interdigital space was sufficiently open, and the skin was not too tense. If necessary, a skin graft or Z-plasty was performed to create the interdigital space. Reconstruction in all patients was performed by the same surgeon.

RESULTS

Over ten years, we had three cases of thumb polydactyly that were treated with on-top plasty. All cases had a normal nail and developed distal part distal to the middle of the proximal phalanx in the ulnar thumb, and their radial thumb was tapered. In two of three cases (Cases 1 and 2), the ulnar thumb was floating and attached to the first web of the patient's hand.

Case presentations

Case 1. A one-year-old boy had radial polydactyly in his right hand. The patient was born after 39 weeks, weighing 3100 g with left-side cryptorchism. The patient had no notable family history and a normal left thumb. The patient's polydactyly consisted of two thumbs (Figure 3). The ulnar thumb floated and adhered to the first web and had a nail, interphalangeal joint, and distal and proximal phalanx. The transverse diameter and thickness of the ulnar thumb were almost equivalent to those of the left thumb. The radial thumb was not floating and showed

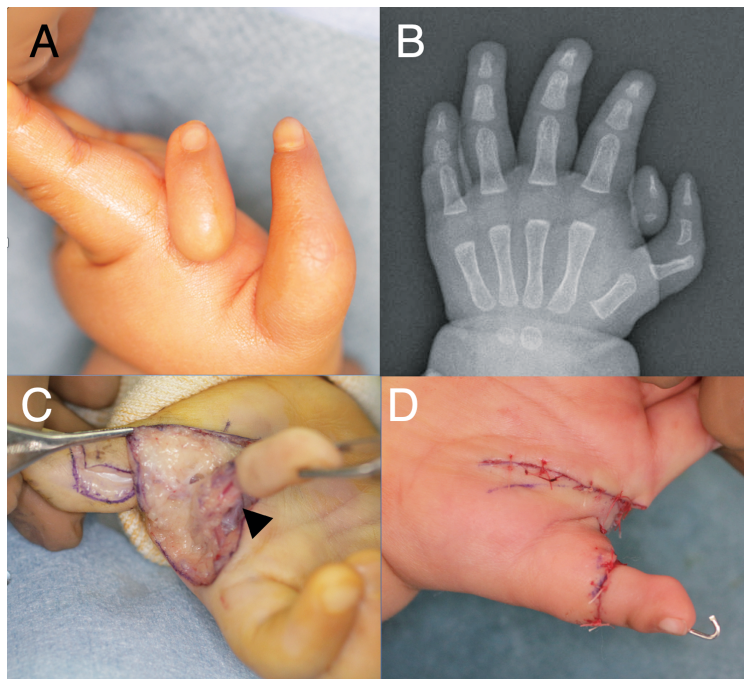


Fig. 3 Case 1: Photographs and radiographs in the perioperative period
Fig. 3A–D: A preoperative photograph (A) and a posterior-anterior radiography image (B). Intraoperative photographs show the neurovascular bundle (arrowhead) included in the flap and appropriately placed on the proximal stump of the dissected radial thumb (C, D).

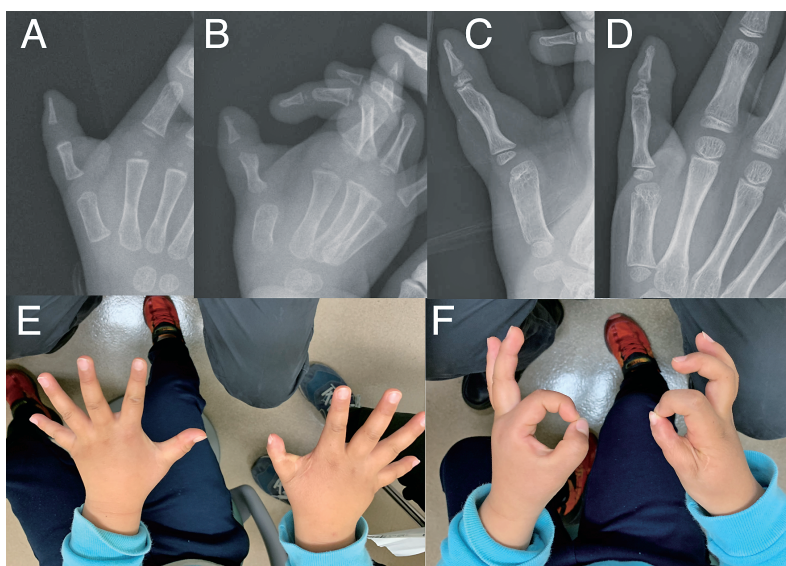


Fig. 4 Case 1: Postoperative photographs and radiographs
Fig. 4A–F: Eight-month (A, B) and six-year (C, D) postoperative radiographs. At the latest follow-up, the patient had a limited range of motion in the metacarpal joint but did not feel any hand function inconvenience (E, F).

triphalangism, was wholly hypoplastic, and the distal end was tapered. The radial thumbnail was also hypoplastic, but the thenar muscle was not. We performed surgery on the thumb when the patient was one year old. The ulnar thumb had normal carpometacarpal and metacarpophalangeal joints. We designed the operation plan using both thumb parts, considering that the ulnar part was not hypoplastic (but floating) and the radial part was hypoplastic.

Four weeks after the operation, the bone union was identified by radiography. The patient accepted being touched but did not try to use the thumb. After two months, the patient began to use the thumb when gripping a toy, and after six months, the patient could hold a pen between the thumb and palmar surface of the index finger for poking. One year after the operation, the patient used the thumb for writing but could not grasp the tips of the thumb and index finger because the interphalangeal joint of the thumb presented lateral instability. Therefore, we performed arthrodesis of the interphalangeal joint. Six years after the first operation, the patient used the thumb without difficulty (Figure 4). There was no pain or numbness in the thumb. The active arc of motion of the metacarpophalangeal joint was 75°, and the interphalangeal joint was ankylosed. The PedsQL score was 100, and the face scale regarding satisfaction was 4.

Case 2. A one-year-old girl had bilateral thumb polydactyly and visited our hospital immediately after birth. The patient was born after 39 weeks weighing 2000 g. The patient had aproctia and underwent proctoplasty, and also had bilateral radial polydactyly. As the left thumb was floating and necrotic at birth, it was resected at the same time as the proctoplasty (Figure 5).

The patient was a candidate for an on-top plasty for polydactyly. The ulnar thumb with a normal nail was floating and adhered to the radial side of the index finger (Figure 5), with an interphalangeal joint and distal and proximal phalanx. The transverse diameter and thickness of the ulnar thumb were almost equivalent to those of the left thumb. The radial thumb was not

floating and had a normal metacarpophalangeal and carpometacarpal joints, but distal phalanx was not present. The polydactyly, similar to Case 1, was not classifiable.

We performed surgery to reconstruct the right thumb when the patient was one year old. We designed a spare part surgery using both parts, similar to Case 1. Four weeks after the operation,

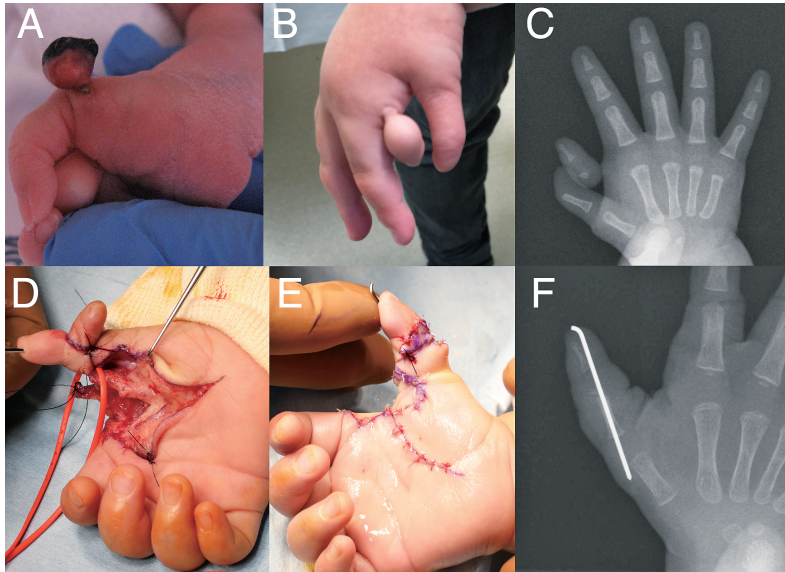


Fig. 5 Case 2: Photographs and radiographs in the preoperative and perioperative period
Fig. 5A–F: Photograph of the left hand (A) just after birth. A right-hand photograph (B) and radiograph before reconstructive surgery (C). On-top plasty was performed with the protection of the ulnar thumb neurovascular bundle (D–F).

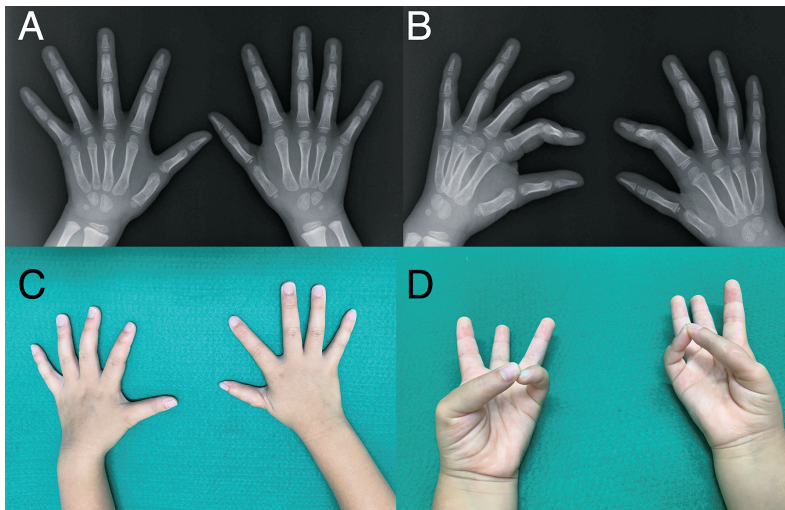


Fig. 6 Case 2: Postoperative photographs and radiographs
 Five years postoperatively, the interphalangeal joint of the right thumb was ankylosed. The range of motion of the thumb is restricted, but there was no trouble using both thumbs.

the bone achieved union, and the patient accepted being touched. After seven months, the patient could hold a pen between the thumb and palmar surface of the index finger for poking. After, we found lateral instability of the interphalangeal joint, and joint arthrodesis was planned. The joint became rigid, but the patient did not suffer from it. Therefore, arthrodesis was not performed. Five years after surgery, the interphalangeal joint was ankylosed. The active range of motion of the metacarpophalangeal joint was limited (30° flexion; 0° extension) but compensated by the carpometacarpal joint motion, and there was no pain or numbness in the patient's thumb. The patient did not feel any thumb function inconvenience (Figure 6). The PedsQL score was 96, and the face scale was 4.

Case 3. A girl with polydactyly accompanied hypoplasia of the thenar muscle first visited our hospital when she was two years old. The patient was born at 40 weeks, weighing 2700 g without complications. The patient had patent ductus arteriosus, but an additional intervention was not required. The patient's left hand was in a normal configuration, and the right hand had radial polydactyly. The radial thumb included the normal metacarpal joint and proximal phalanx, but it tapered and lacked the distal phalanx and nail. The ulnar thumb had a normal nail and a distal phalanx (Figure 7). Additionally, the thenar muscles were hypoplastic. Thus, we planned plastics of the thumb tip with on-top plasty followed by an opponenseplasty.

Tip reconstruction was performed in the same way as in the previous two cases when the patient was two years old. The ulnar thumb was placed on the radial thumb without rotation. However, before the bone fusion, the patient accidentally removed the Kirchner wire, and the distal part rotated 30° . Thus, one year after the surgery, an osteotomy was performed in the metacarpal to correct the rotation. Three years postoperatively, the correction of the rotation of the thumb was not complete; however, the patient and his family did not wish to proceed with further reconstruction. The arc of the metacarpophalangeal joint was 45° , and the interphalangeal joint hardly actively moved. The patient's thumb had no pain or numbness. Subsequent improve-

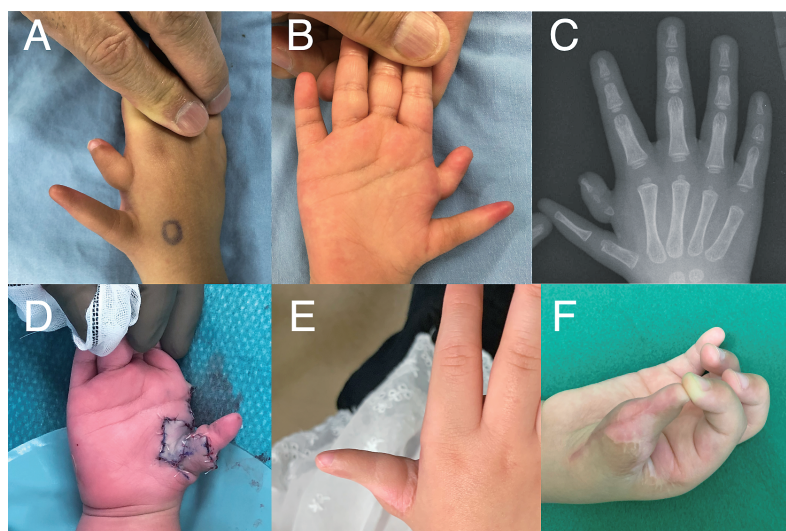


Fig. 7 Case 3: Photographs and radiographs in the preoperative, intraoperative, and postoperative periods
Fig. 7A–F: Preoperative photographs (A, B) and a radiography image (C). The on-top plasty was performed with a skin graft (D). A photograph of the rotated distal part preoperatively (E) and after osteotomy (F).

ment in the atrophic, hypoplastic thenar muscle allowed oppositional movement of the thumb. The latest PedsQL score was 96, and the face scale score was 3. The patient did not experience any difficulty in the use of the thumb.

Literature review

We found 11 studies that described cases of on-top plasty for thumb polydactyly (Table 1, 2).^{1,2,4-10,14,15} One study that included an on-top plasty case was excluded because it did not

Table 1 Reported cases of radial polydactyly treated with on-top plasty: patient demographics

Authors and publication year	Number of reported cases	Sex	*Patient age	*Follow-up period	
Dobyns JH et al ¹⁴	1985	1	N/A	N/A	10
Yen CH et al ¹⁵	2006	1	male	2.3	3
Hasegawa K et al ¹	2013	1	male	1	0.6
Iba K et al ²	2013	2	N/A	1.2	1
Al-Qattan NM et al ⁴	2017	1	male	5	10
Bell B et al ⁵	2017	5	4 males, 1 female	1.4	13.5
Kelly BP et al ⁶	2018	1	male	1.7	N/A
Pons-Riverola A ⁷	2018	1	male	1.2	10
Hara T et al ⁸	2019	1	female	3.5	15
Zhong W et al ⁹	2020	3	2 males, 1 female	2.6	N/A
Fang X et al ¹⁰	2021	21	14 males, 7 females	1.2	5.1

* For multiple cases, the average values are reported.

Table 2 Reported cases of radial polydactyly treated with on-top plasty: Functions of the reconstructed thumbs

Authors	*Arc of MPJ and IPJ	Other described functions and comments
Dobyns JH et al ¹⁴	N/A	MPJ instability and weak opposition power required secondary reconstruction.
Yen CH et al ¹⁵	N/A	
Hasegawa K et al ¹	N/A	Bony union between the graft and recipient was not identified; thumb instability did not occur.
Iba K et al ²	N/A	Both were Wassel type 6 and the graft included MPJ and IPJ.
Al-Qattan NM et al ⁴	60,60	
Bell B et al ⁵	60,19	PedsQL for parents 89, DASH 4.3
Kelly BP et al ⁶	N/A	Stiff IPJ, but the thumb was opposable to all four digits.
Pons-Riverola A ⁷	N/A	Pinch power: 3 kgf (contralateral side: 4 kgf)
Hara T et al ⁸	60,0	Arthrodesis was added eight year after the first surgery.
Zhong W et al ⁹	N/A	
Fang X et al ¹⁰	75,43	PedsQL; for child = 85, for parents = 92.

MPJ: metacarpophalangeal joint

IPJ: interphalangeal joint

PedsQL: Pediatric Quality of Life Inventory

DASH: disability of the arm, shoulder, and hand

* For multiple cases, the average values are reported.

describe the therapeutic process.¹⁶ Most studies included one case. However, one study included 21 patients who underwent surgery over ten years. There was a slight tendency for boys to be indicated for surgery more than girls. As in our cases, the most frequent polydactyly pattern was a developed proximal part of the radial thumb and a developed distal part, including the nail. In this pattern, the ulnar thumb is sometimes floating, and a defect is proximal to the middle of the proximal phalanx.

Patients underwent reconstruction by transplanting the graft, including the interphalangeal joint at the top of the proximal part. Iba et al reported a special transfer, including the metacarpophalangeal joint for the Wassel type 6 polydactyly.² Fang et al reported that the graft level was determined based on the degree of metacarpal joint development.¹⁰ Most patients underwent surgery from one to two years of age when children usually start grasping, using their thumb. In two cases, patients underwent reconstruction when they were three and four years old.^{4,8} Most surgeons performed surgery at one time, and two authors reported operations in two separate procedures to prevent congestion of the flap.^{1,9}

At the final follow-up, all authors reported good appearances, clearly illustrated in the published images. Table 2 also summarizes the thumb functions. Some reports only considered a good appearance as a success and did not describe function. The range of motion in the metacarpophalangeal and interphalangeal joints demonstrated a good arc of the metacarpophalangeal joint. Furthermore, Al-Qattan et al and Fang et al reported that the average arc in the interphalangeal joint was 50° and 43°, respectively,^{4,10} but they were not in the active range of motions.

Joint instability after reconstruction was reported by Hara et al, and arthrodesis was performed eight years postoperatively for severe interphalangeal joint instability.⁸ Dobyns et al reported instability of the metaphalangeal joint, which was reconstructed in the secondary surgery.¹⁴ Other published cases did not report additional surgeries for unexpected complications.

Three authors reported the function using patient-related outcomes, such as PedsQL.^{5,8,10} The outcome and satisfaction evaluation measurements were different in each study, but, in all cases, the patients and their parents were satisfied with the thumb function.

DISCUSSION

Surgeons are sometimes required to design special operation plans to treat polydactyly because there is no apparent dominant thumb. On-top plasty is an atypical reconstruction of thumb polydactyly, estimated to be performed in less than 1 % of all thumb reconstructions.¹⁷ Cases with a thumb with a normal configuration of the distal part, including the nail and another thumb with normal development in the proximal part, are good indications for using on-top plasty. In the procedure, the developed distal part of one thumb was placed on top of the proximal part of the other thumb. By using this scheme, we can reconstruct the thumb with a good appearance.

We assumed that the patients began using the thumb in the early postoperative period because the sensory and appearance of the thumb were reconstructed as a normal thumb. However, patients did not use the thumb for more than two months postoperatively. Thus, the tension on the nerves might have caused transient hyposensitivity or even hypersensitivity.

We encountered interphalangeal joint instability in two cases (patient 1 and 2). As thumb joint stability is important for acquiring thumb function, we conducted arthrodesis of the interphalangeal joint before the patient learned the tip pinch. One patient (patient 1) required arthrodesis. Although the joint was reconstructed with a vascularized graft, none of the patients could have functional joints. Considering that instability was not evident before the surgery, the blood flow to the joint may not be sufficient to mature the joint. Furthermore, 2 cases (patient 1 and 3)

showed slight atrophy of the fingertip compared to immediately after surgery. In the perioperative period, the blood flow in the flaps was stable without ischemia or congestion. The fingertips did not show any symptoms of atrophy in the short term. The artery in the pedicle and angiogenesis to the flap occurring in the process of the engraftment might contribute to fingertip circulation, but these may not supply enough blood flow for the growth of the fingertip as well as the joint.

We also reviewed published case series or reports of on-top plasty for thumb polydactyly to demonstrate possible joint dysfunction after the procedure. Patients can gain a good and satisfying appearance. However, except for several “best-case scenarios” reported in two studies, the interphalangeal joint motion was restricted, sometimes requiring joint stabilization. Even though the joint anatomy was judged as normal during surgery, the transferred joint cannot always be expected to function as anticipated and may sometimes become rigid or unstable. When performing the on-top plasty for polydactyly treatment, surgeons should inform the patients’ families about the possible joint instability in the graft, which might require additional surgery, such as arthrodesis.

This study has two limitations. First, we evaluated the treatment based on limited information and objective findings. We heard from the patients’ parents about the practical use of the thumb, the hand use was demonstrated in the hospital, and we evaluated a few objective findings. We used PedsEQ as a patient-related outcome, and it was not specified for the follow-up of the reconstruction of congenital deformities. Therefore, it is probable that we were unable to accurately quantify the thumb function, and all the patients should be monitored until they become adults to check for any trouble. Second, the review of reported cases has reporting bias. We assumed that there were more thumb polydactyly cases treated with on-top plasty than previously reported, but negative surgery results were not reported.

On-top plasty for polydactyly is a better procedure than ablation of one thumb regarding the thumb’s appearance. However, the patient rarely gains a functional interphalangeal joint, and joint instability is possible, which are disadvantages. These dysfunctions are not major problems and are likely outweighed by patient satisfaction and appearance.

ACKNOWLEDGEMENT

This work was supported by the Grants-in-Aid for Scientific Research of Japan Society for the Promotion of Science (Grant Number JP21H03288, Hidemasa Yoneda). We would like to thank Editage (www.editage.com) for English language editing.

CONFLICT OF INTEREST

None.

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