

# Clinical Characteristics and Surgical Outcomes of Congenital Ulnar-deviated Thumbs: Delta Triphalangeal Thumbs and Irregular Epiphyses

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**Background:** Delta triphalangeal thumbs (DTPT) and irregular epiphysis thumbs (IET) had different anatomic deformities. Our primary purpose was to evaluate the clinical and radiographic outcomes of surgical treatment in DTPT and IET.

**Methods:** In total, 43 ulnar-deviated thumbs were included and categorized into 2 types according to x-ray and exploration during surgery, DTPT and IET. Surgical excision of the delta phalanx in DTPT and intraepiphysis osteotomy in IET was conducted.

**Results:** In total, 23 ulnar-deviated thumbs were classified as DTPT and 20 as IET. Ten thumbs that could not be classified initially were followed-up until they could be categorized at the mean age of 24 months. The preoperative mean degrees of ulnar deviation at the interphalangeal joints were 40 and 33 degrees, in DTPT and IET, respectively. The mean degrees were 2 and 5 degrees in final follow-up, showing significant improvement (DTPT,  $P < 0.05$ ; IET,  $P < 0.05$ ). Complications during the study included residual ulnar deviation, overcorrection, and nonunion. The stability and range of movement at the interphalangeal joint were good overall. According to the Japanese Society for Surgery of the Hand scoring system, results were excellent in 29 cases, good in 13, and fair in 1.

**Conclusions:** Ulnar clinodactyly of the thumb occurs because of different anatomic features such as DTPT or IET. We recommend surgical treatment be postponed until the anatomic abnormality can be ascertained. Furthermore, almost all patients with ulnar-deviated thumbs had significant improvement in clinical and radiographic outcomes after surgery.

**Key Words:** clinodactyly, delta thumb, triphalangeal thumb, delta epiphysis, surgery

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The clinical definition of ulnar deviation of the thumb shows abnormality of the interphalangeal (IP) joint causing the thumb to be misaligned and point toward the fifth finger. Furthermore, ulnar deviation of the thumb often presents as an isolated anomaly or part of the thumb with other congenital malformations of the hands.<sup>1,2</sup> Most ulnar-deviated thumbs are categorized as delta triphalangeal thumbs (DTPT).<sup>3–5</sup> However, when Ogino et al<sup>6</sup> presented a classification system of the opposable triphalangeal thumb (TPT) in 1994; and they reported that the accessory phalanx was triangular in shape and resembled a secondary ossification center of the distal phalanx. They were the first to propose the concept of an abnormal ossification center. Baek et al<sup>7</sup> treated 6 patients with abnormal triangular epiphyses in 2006. Both these assumptions about abnormal epiphyses are based on x-ray image detection. We present different pathologic characteristics in DTPTs and irregular epiphysis thumbs (IET) on the basis of x-ray, exploration during surgery, magnetic resonance imaging, and pathologic examinations.<sup>8</sup> However, there are few studies in the literature focusing on the outcomes of surgical treatment between them. As we known, it was the first study to report the clinical and radiographic outcomes in DTPT and IET.

## METHODS

Clinical data of 36 patients with 52 ulnar-deviated thumbs treated surgically in our hospital between September 2011 and December 2016 were retrospectively reviewed. Patients with nonopposable triphalangism; a TPT with other associated abnormalities, except for radial polydactyly; and duration of follow-up of <24 months were excluded from this study. A total of 43 ulnar-deviated thumbs with 25 preaxial polydactyly (17 patients) in 28 patients were included. All patients were treated by a single surgeon (Y.X.). Before the surgery, the clinical data of patients including sex, age, affected side, and associated anomalies were recorded. Range of motion was measured with a goniometer preoperatively and reassessed during the final follow-up. If the patients were younger than 3 years old before surgery, range of motion was measured passively—the other age groups were measured actively instead. At the final follow-up, all were measured actively. Alignment at IP joints were measured on the preoperative and postoperative x-ray films. The Japanese Society for Surgery of the Hand score system (JSSH)<sup>9</sup> evaluation form was adopted to assess the functional and cosmetic outcomes of the reconstructed thumbs.

All cases were categorized into 2 types, DTPT and IET, on the basis of the roentgenographic findings. If there were unclear decisions according to the x-ray presentation in patients with extremely underdeveloped skeletons, the observation was recommended to be followed-up every 3 to 6 months by x-ray film, until classification could be ascertained, and further confirmed during the surgery. In total, 11 thumbs were confirmed as IET and 8 thumbs could not be classified initially, and they were recommended to undergo surgical resection of duplicated thumbs, the surgical correction of the ulnar-deviated thumbs were postponed.

The surgical strategies vary according to anatomic characteristics. Excision of delta phalanx was undergone in DTPT case and intraepiphysis osteotomy in IET cases. Under tourniquet control, either a longitudinal radial median incision was made at the IP joint on the convex side of IET or a radial transverse fusiform incision in DTPT. Fluoroscopy was used to confirm the location of the irregular epiphyses or delta phalanx. Excision of the delta phalanx in DTPT (Fig. 1E), and reparation of the collateral ligament using local capsular tissue, was completed with a retrograde longitudinal 0.048-inch Kirschner wire fixation (Double Medical Technology Inc, Xiamen City, China) (Fig. 1F).

Intraepiphysis osteotomy is the critical technique in IET, the proximal osteotomy line was located in parallel with the IP articular surface and the distal osteotomy line was located in parallel with epiphyseal plate, respectively (Fig. 2E). The closing wedge osteotomy was performed by a blade. The angulation of the thumb was then corrected to a neutral alignment. A retrograde longitudinal 0.048-inch Kirschner wire was then advanced across the osteotomy and IP (Fig. 2F).

The affected thumbs were immobilized with long-arm cast. The Kirschner wires were removed 3 to 4 weeks with DTPT and 4 to 6 weeks with IET after surgery, especially healing was confirmed radiographically in osteotomy cases.

Statistical significance was set at  $P < 0.05$ , and it was determined with a 2-tailed Student *t* test. Informed consent was obtained and institutional review board approval was acquired.

## RESULTS

In total, 28 patients (10 male and 18 female) were included in the initial assessment. The right side was affected in 9 patients, the left side in 4 patients, and both

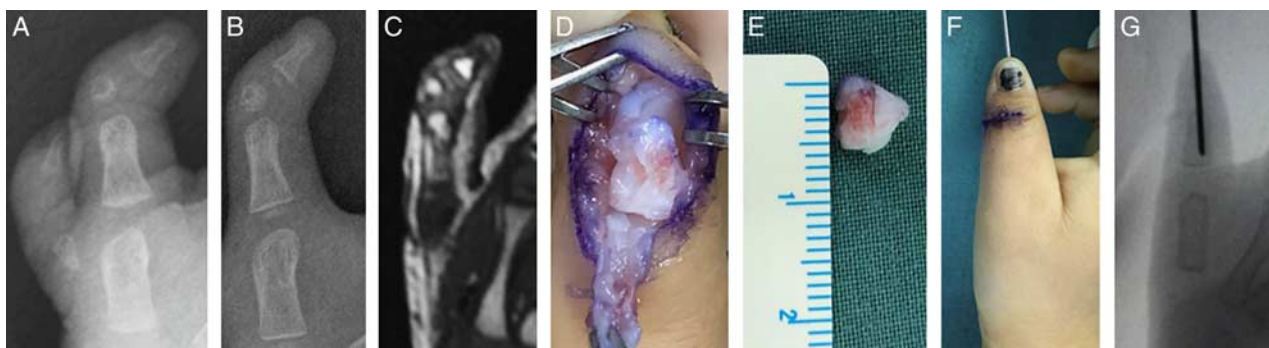
sides in 15 patients. Of the 43 ulnar-deviated thumbs, 23 (53.5%) were classified as DTPT and 20 (46.5%) were as IET. Of the 15 bilateral cases, 7 patients had DTPT in 1 hand and an IET on the contralateral side. The mean age at the first examination was 12 months old (range, 6 to 23 mo). There were 17 patients (25 thumbs, 58%) with a duplicated thumb, and 6 of these thumbs were initially confirmed as DTPT and underwent 1-stage resection of duplicated thumb and accessory phalanx.

In total, 10 thumbs (includes 8 thumbs with duplicated thumb) with a mean age of 8 months, who could not be classified initially, were followed-up and observed in the clinic until they could be categorized at the mean age of 24 months (range, 16 to 30 mo). Seven of the 10 were categorized with DTPT on the basis of x-ray and 3 of the 10 with IET (Fig. 3).

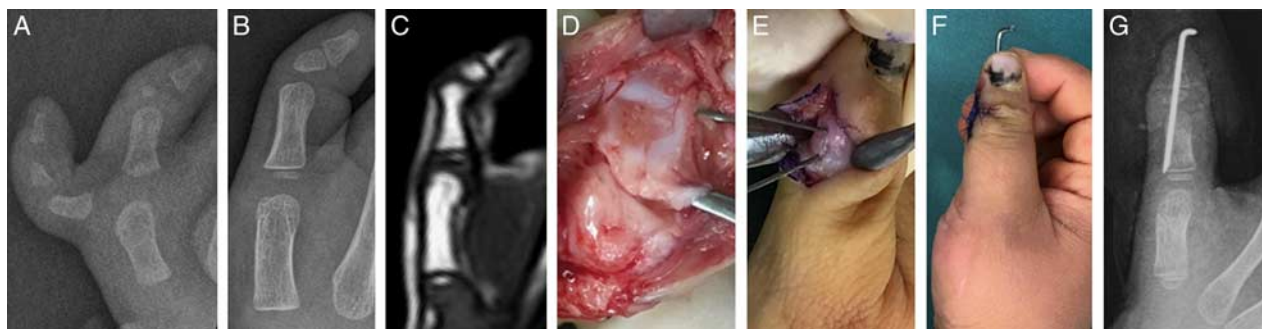
It was difficult to distinguish irregular epiphyses of distal phalanxes from conventional DTPTs with the absence of epiphyses of distal phalanxes. A right ulnar-deviated thumb with radial polydactyly showed unclear presentation using an initial x-ray presentation in a 9-month-old boy (Fig. 2A). Postoperation of thumb duplication, he was confirmed with the diagnoses as an IET until he was 30 months old (Fig. 2B). Another 8-month-old boy with a right ulnar-deviated thumb and radial polydactyly underwent polydactyly resection. It was unclear whether to make a decision according to the initial early x-ray presentation (Fig. 1A). DTPT was ascertained when the patient was 23 months old, exhibiting epiphysis of the distal phalanx demonstrated, through x-ray (Fig. 1B).

The mean ages at the surgery of DTPT were 16 months (range, 7 to 23 mo) and 33 months (range, 19 to 48 mo) in IET, respectively. The mean follow-up evaluation in our cases was 39 months (range, 24 to 70 mo). The alignment of the IP joints was significantly improved at the final follow-up in all cases. The stability of the IP joint was good overall (Fig. 4).

The mean value of ulnar deviation at the IP joints in DTPT decreased from 38 to 2 degrees postoperatively ( $P < 0.05$ ), respectively. Preoperative flexion of IP joints averaged 53 degrees and the mean value was 43 degrees at the final follow-up ( $P > 0.05$ ). The mean extension lag was



**FIGURE 1.** A–G, A 8-month-old boy, right ulnar-deviated thumb with radial polydactyly, it was ambiguous on x-ray image. He was confirmed as a delta triphalangeal thumbs at 23 months old and underwent excision of the delta phalanx later. full color online



**FIGURE 2.** A–G, A 9-month-old boy, right ulnar-deviated thumb with radial polydactyly, it was ambiguous on x-ray presentation, with surgical resection of duplicated thumbs first. He was confirmed as an irregular epiphysis thumbs at the age of 30 months, underwent closing wedge osteotomy. [full color online](#)

16 degrees (ranging from 8 to 23 degrees) at the IP joint preoperatively; their mean values increased to extension 5 degrees at the final follow-up ( $P < 0.05$ ).

The mean ulnar deviated at the IP joints in IET cases decreased from 35 to 5 degrees postoperatively ( $P < 0.05$ ). Preoperative flexion of IP joints averaged 47 degrees and the mean value was 43 degrees at the final follow-up ( $P > 0.05$ ). The mean extension lag was 13 degrees (ranging from 10 to 22 degrees) at the IP joint preoperatively; their mean values increased to extension 3 degrees postoperatively ( $P < 0.05$ ) (Table 1).

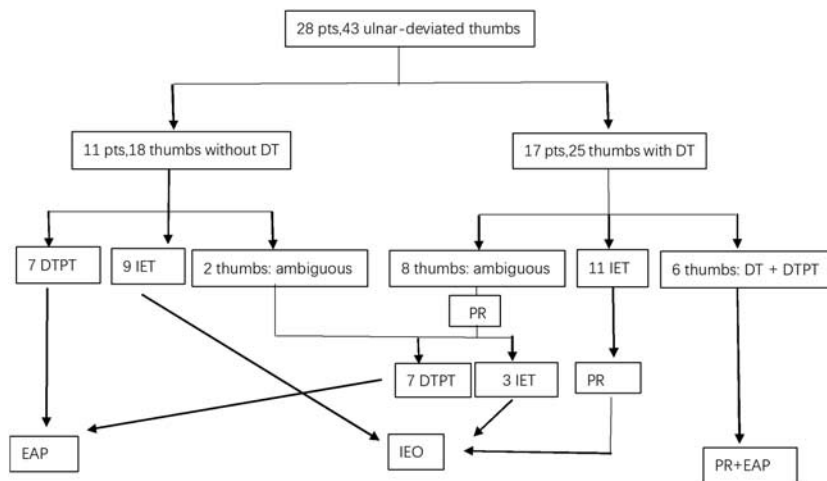
According to the JSSH scoring system, the results were excellent in 29 cases, good in 13, and fair in 1. In the IET group, 1 patient exhibited 47 degrees ulnar deviation, at the IP joint preoperation, then displayed 23 degrees of residual ulnar deviation after 4 years postsurgery. This patient underwent a revised closing wedge osteotomy of the abnormal epiphyses. Nonunion was observed radiographically in 1 case 2 years after intraepiphysis osteotomy, although there were no signs of discomfort or growth disorder, in the most recent follow-up; thus, long-term evaluation is recommended.

Besides, 1 case with 12 degrees overcorrection in DTPT is still being evaluated.

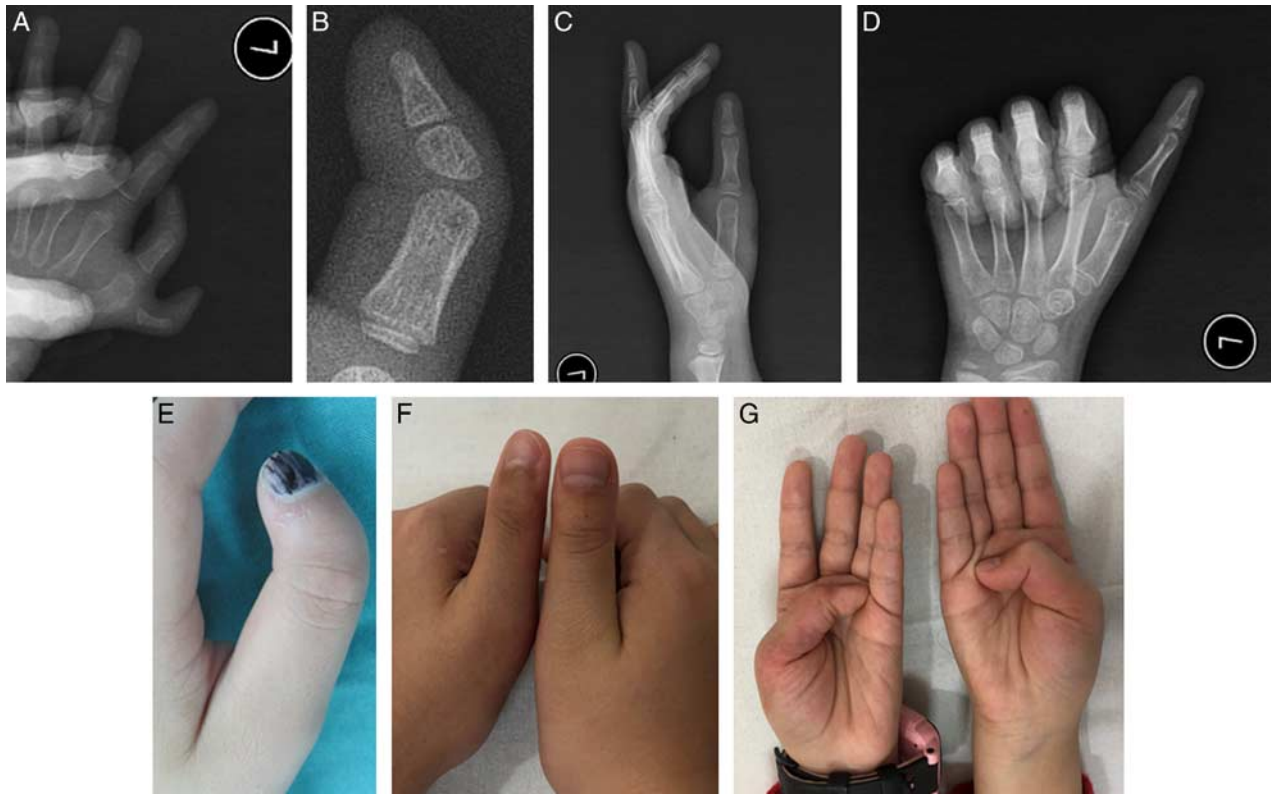
No patient reported pain or difficulties in daily living. All parents were satisfied with the appearance of affected thumbs except for the revised case. In the epiphyseal deformity cases, 1 case was followed-up for 6 years after surgery, and the epiphysis of the distal phalanx was because of premature closure (Figs. 4C, D).

### DISCUSSION

Traditionally, most ulnar-deviated thumbs were categorized as TPTs, which was rare, occurring at an incidence of roughly 1 in 25,000.<sup>3</sup> These cases represent ~3% of upper-extremity congenital anomalies.<sup>4</sup> Ogino et al<sup>6</sup> presented a classification system of the opposable TPT in 1994; in their type 2 thumbs. They showed that the accessory phalanx was triangular and resembled a secondary ossification center of the epiphysis of the distal phalanx; however, they did not affirm the accurate structure by other methods aside from x-ray films. We had determined



**FIGURE 3.** Diagnostic algorithms of 43 ulnar-deviated thumbs in our study. DT indicates duplicated thumb; DTPT, delta triphalangeal thumbs; EAP, excision of accessory phalanx; IEO, intraepiphysis osteotomy; IET, irregular epiphysis thumbs; PR, polydactyly resection.



**FIGURE 4.** A and B, A 12-month-old girl, left ulnar-deviated thumb with radial polydactyly, it was ambiguous on x-ray image. She was confirmed as a irregular epiphysis thumbs at 21 months old. C and D, At 6 years after intraepiphysis osteotomy operation, the x-film indicated no deviation of interphalangeal joint, epiphyseal morphology was normal, but the epiphysis of distal phalanx tended to be premature closure. E, Appearance before the operation. F and G, Satisfied appearance and function at 6 years after the operation. full color online

that IET do not involve a triphalanx digit, and it should not be categorized as TPT. Thus, ulnar clinodactyly of the thumb occurs because of 2 types of anatomic features, DTPT or IET, which was described in our previous report.<sup>8</sup> In our study, 10 thumbs of patients with a mean age of 8 months, with the small ossicular located between the phalanxes could not be classified initially; at the mean age of 24 months, 7 of the 10 thumbs were subtype 1A and categorized as DTPT, and 3 of the 10 thumbs were subtype 1B and categorized as the IET.

Although DTPTs are relatively simple Wood<sup>4</sup> type I-TPT, surgical treatment of the delta accessory phalanx has been controversial. The first approach for young patients is based on excision of the extra ossicular, with or without

reconstruction of the collateral ligament and pinning. This is typically recommended for joint remodeling of the new IP joint with time allowed in young patients.<sup>4,10-14</sup> Thus, we adopted simple excision of the delta phalanx with pinning and collateral ligament reconstruction in this series in patients who presented with ulnar-deviated thumbs with extra delta phalanxes.

In the surgical treatment of IET, Ogino et al<sup>6</sup> described a groove between the original distal phalanx and “extra phalanx.” The proximal part of the groove, which included a part of articular cartilage and “extra phalanx,” was removed. Partial excision of the epiphysis was also performed by Horii et al<sup>15</sup> for 11 trapezoidal epiphyses; however, they found that the groove was not clear, and it was difficult to determine the amount of epiphysis that

**TABLE 1.** Preoperative and Postoperative ROM Plus Alignment Data

	IP in DTPT			IP in IET		
	ROM (deg.)		Alignment	ROM (deg.)		Alignment
	Extension	Flexion		Extension	Flexion	
Preoperative	-16 ± 2	53 ± 5	38 ± 2	-13 ± 2	47 ± 4	35 ± 2
Postoperative	5 ± 2	43 ± 3	2 ± 0.5	3 ± 1	43 ± 3	5 ± 1
<i>P</i>	0.014	0.36	<0.01	0.023	0.27	<0.01

DTPT indicates delta triphalangeal thumbs; IET, irregular epiphysis thumbs; IP, interphalangeal joint; ROM, range of motion.

could be excised. Baek et al<sup>7</sup> treated 6 patients with abnormal triangular epiphyses in the distal phalanx of the thumb, including 3 bilateral cases. They suggested that abnormal triangular epiphyses can be treated with intraepiphyseal closing wedge osteotomy. We believe that IET have 2 phalanxes and should not belong to the subtype of TPTs; therefore, we recommended intraepiphyseal-corrective osteotomy to retain both the epiphyseal plate and articular cartilage of the distal phalanx. Alignment deviation after operation at the IP joints was reported by Horii et al.<sup>15</sup> In our IET series cases, almost all patients with ulnar-deviated thumbs had significant improvement in clinical and radiographic outcome after surgery, and only 1/20 recurred 23 degrees of residual ulnar deviation at the IP joint 4 year after surgery and accepted revised intraepiphyseal osteotomy. At the final follow-up, all patients were satisfied with appearance and function.

Well-defined distal phalangeal epiphyses of the thumb are usually not clearly evident until male infants are  $20.6 \pm 6.8$  months old and female infants are  $13.2 \pm 5.5$  months old.<sup>16</sup> Occasionally, it seems to be difficult to qualitatively analyze small ossicular that are located between the phalanxes on x-ray before the age of 1 year; subsequently, close observation and periodic follow-up evaluation are reasonable, the surgical treatment were postponed until the anatomic characteristics could be categorized at the mean age of 24 months, which all were less than 30 months. Early surgery for the delta phalanx makes sense, but a bit of delay to best understand the irregular epiphysis, and also give it time to sufficiently enlarge for satisfactory surgery makes sense.

In DTPT cases, excision of the delta phalanx in DTPT (Fig. 1E). After removing the delta phalanx, we repaired the collateral ligament using local capsular tissue, with proper tension (not too tight or too loose) and completed them with a retrograde longitudinal Kirschner wire fixation in neutral position (Fig. 1F). In IET cases, the proximal osteotomy line was located in parallel with the IP articular surface and the distal osteotomy line was located in parallel with the epiphyseal plate, respectively. The periosteum should be protected entirely. The osteotomy does not involve the articular surface and was finished by blades. Kirschner wire was used to fix in neutral position. Do not pass the Kirschner wire multiple times through the bone.

Our study is limited by the retrospective design, absence of control group, and the small number of patients because of the extremely low incidence of ulnar-deviated thumbs. JSSH scoring was influenced by the factors of the duplicate thumbs; in addition, our mean follow-up of 39 months is relatively short because of the potential growth disturbance.

In conclusion, ulnar clinodactyly of the thumb is because of different anatomic features, DTPT or IET, and it is closely related to surgical strategies. The surgical treatment should be postponed when the radiographic presentation is ambiguous until the anatomic abnormality can be ascertained by imaging examination. Excision of the delta phalanx in DTPT and intraepiphysis osteotomy in IET were recommended with significant improvement in clinical and radiographic outcomes.

Because the osteotomy was located in intraepiphysis, patients have substantial growth potential. Longer-term follow-up until bone maturity will be important for the assessment of the effects of our treatment on growth.

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