

# Polydactyly and the triphalangeal thumb

*Polydactyly associated with triphalangeal thumb is not rare, as among the records of 1,269 patients with congenital deformation of the upper extremity at the University of Iowa there were 21 patients with 32 hands with such involvement. More detailed classification based on Wassel's work was made. Treatment should be early, with removal of the radial digit if equal in size, excision of all accessory parts including delta bone when present, reconstruction of joint ligaments or joint fusion and osteotomy for correction of deviations. Care should be taken to avoid leaving a narrow first web space.*

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The triphalangeal thumb often has been recorded in association with polydactyly.<sup>1-3</sup> Even though most authorities consider this to be a rare congenital malformation,<sup>4-6</sup> it is probably not as rare as has been implied.<sup>7</sup>

We have records of 42 patients with a total of 68 triphalangeal thumbs and of 203 patients with polydactyly of 286 digits. Twenty-one of these patients have an associated thumb polydactyly with triphalangism in 32 hands. These are part of a group of records of 1,269 patients collected at the University of Iowa who had congenital abnormalities of the upper extremity. The association of polydactyly with a triphalangeal thumb represents an incidence of about 2% of congenital abnormalities of the upper extremity in our records.

Several reports describe the association of these two anomalies.<sup>8-11</sup> Of Dubreuil-Chambardel's<sup>2</sup> 74 cases of triphalangeal thumb, 32 were associated with preaxial polydactyly.

## Inheritance

This particular combination of deformities appears to be inherited as a simple autosomal dominant.<sup>12, 13</sup> Ten of our 21 patients had a family history of the same abnormality in close relatives.

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**Table I.** Thumb polydactyly and triphalangism in 21 patients

Type	No.
Type I	1
Type II	1
Type III	1
Type IV	8
Type V	0
Type VI	0
Type VII	21
Total No. of digits	32

## Clinical findings

There were nine men and 12 women with this combination of malformations. Fifteen had bilateral involvement. Of the patients with unilateral involvement, five patients had only the right hand malformed and only one had the abnormality in the left hand.

## Embryology

Geneticists and anatomists<sup>14, 15</sup> frequently have speculated whether the extra phalanx in triphalangism is a proximal phalanx or a metacarpal. Lapidus, Guidotti and Colleti<sup>16, 17</sup> believe that, in triphalangism, the additional phalanx is not a true middle phalanx but a remnant of the base of one of the phalanges of the bifid thumb. However, many examples have been recorded in which triphalangism and polydactyly have not been associated.<sup>7</sup>

McMurtry<sup>18</sup> developed a technique of cutting serial sections of fetal hands to examine the normal anatomy. Serial sections on an 8-week fetal hand showed the

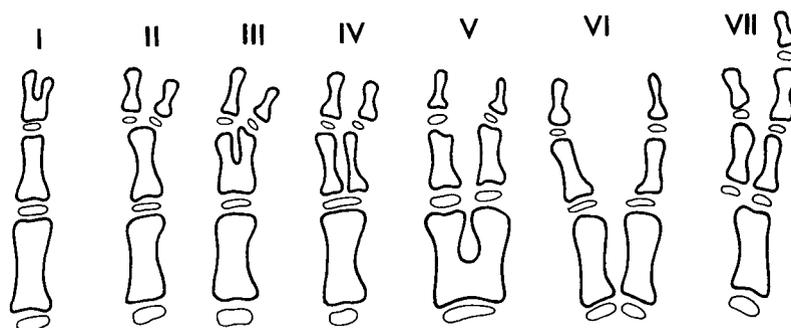


Fig. 1. Wassel's classification of thumb polydactyly.

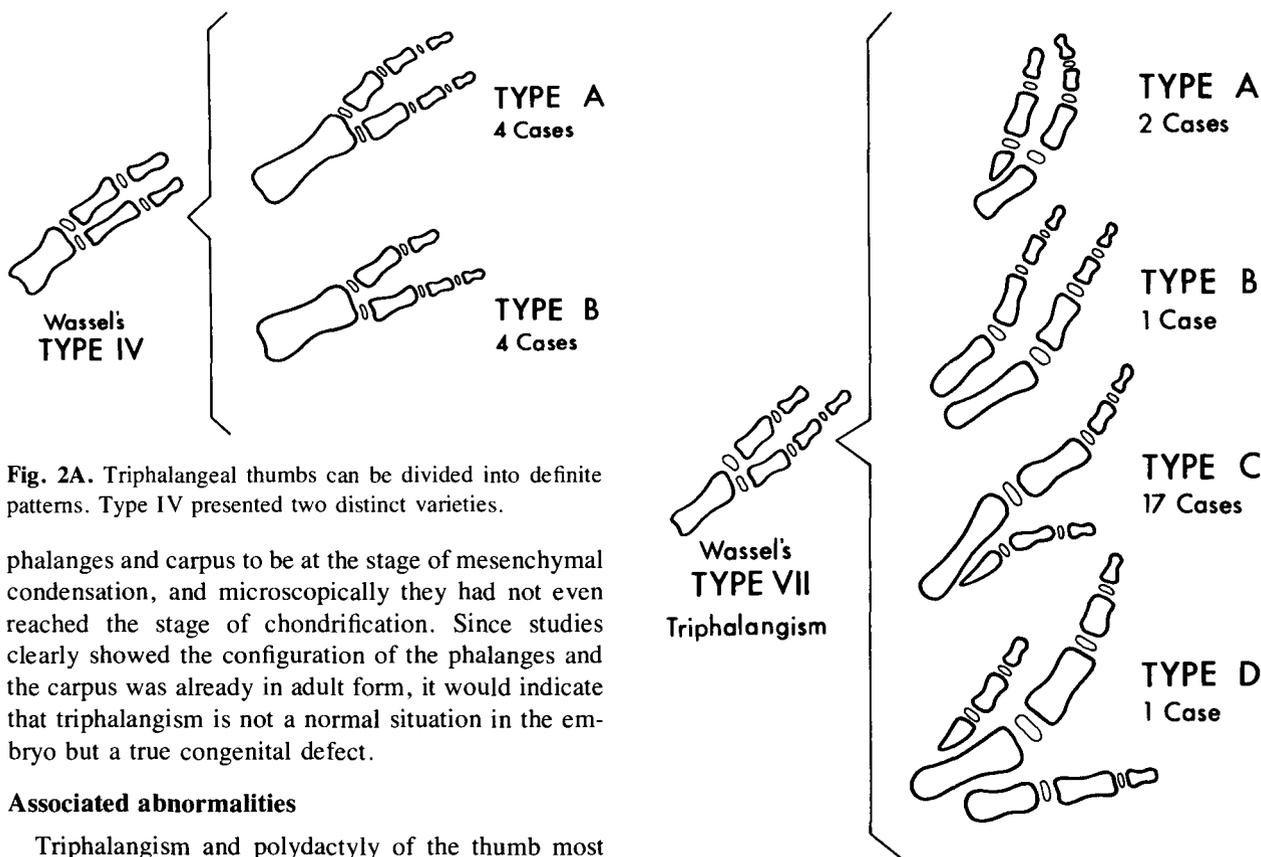


Fig. 2A. Triphalangeal thumbs can be divided into definite patterns. Type IV presented two distinct varieties.

phalanges and carpus to be at the stage of mesenchymal condensation, and microscopically they had not even reached the stage of chondrification. Since studies clearly showed the configuration of the phalanges and the carpus was already in adult form, it would indicate that triphalangism is not a normal situation in the embryo but a true congenital defect.

**Associated abnormalities**

Triphalangism and polydactyly of the thumb most often is associated with polydactyly of the big toe.<sup>6, 19-26</sup> Only two of our patients had polydactyly of the great toe. The only other commonly associated abnormality in our patients with polydactyly and the triphalangeal thumb was the presence of a delta phalanx. Nine of our patients has an associated delta bone.

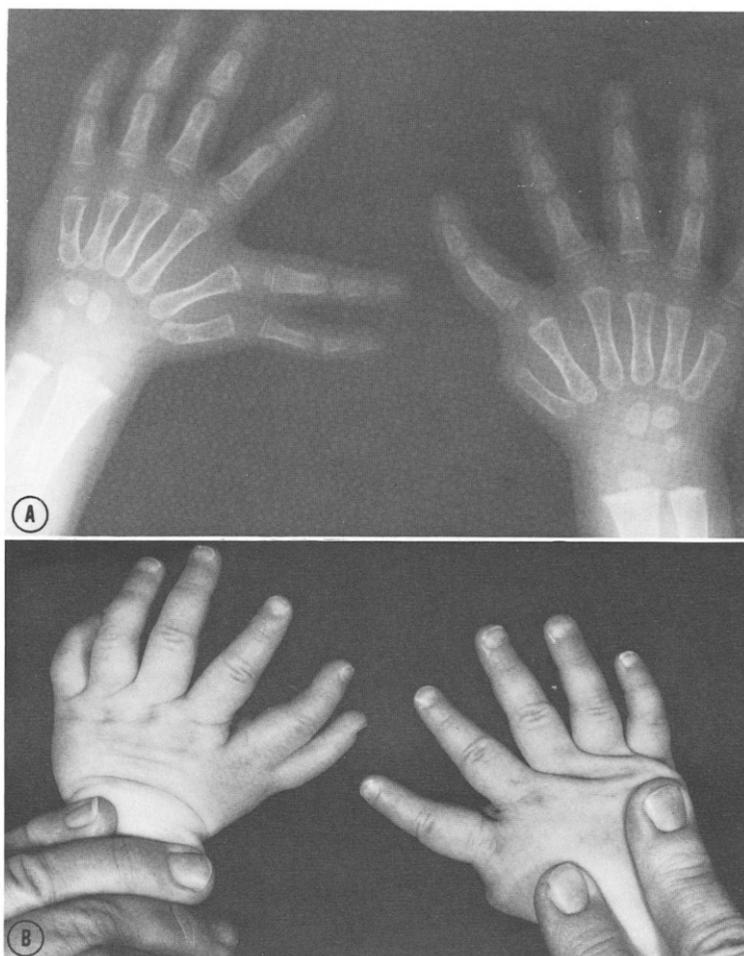
**Classification of the abnormality**

The classification of polydactyly by Wassel<sup>27</sup> was used to divide the patients into seven groups, as shown in Fig. 1. Table I shows the distribution of these seven groups. Twenty-one patients were found with poly-

Fig. 2B. Wassel's type VII rarely occurred. The distribution of our 21 cases of type VII is shown.

dactyly of the thumb in association with triphalangism. These 21 patients had a total of 32 thumbs in which this combination was present.

Type IV polydactyly could be further subdivided into type IV-A and -B (Fig. 2A). Miura<sup>28</sup> reports having one case in which the triphalangeal thumb was on the ulnar side. This could be called a type IV-C polydactyly. Type VII was further divided into four definite patterns. Fig. 2B shows the way these patterns



**Fig. 3.** Different clinical phenotypes (**A** and **B**) are seen of a Type VII polydactyly in the same patient. (From Wood VE: Treatment of the triphalangeal thumb. *Clin Orthop* 120:188-200, 1976.)

are formed. Both the thumb and the supernumerary digit may possess three phalanges as shown in Fig. 3A. The triphalangeal digit may be placed ulnarly or, rarely, it may be on the radial side (Fig. 4, B). More rarely still, it may be duplicated on both sides of the so-called "normal thumb," as shown in Fig. 5, A. These patterns have never been described precisely and grouped. The most frequent pattern seen is type C, as shown in Fig. 4, A. Type VII polydactyly is improper and it should be described more accurately for study purposes, as shown in Fig. 2 B. Tuch et al.<sup>29</sup> avoid classifying these abnormalities and put them all into the category of "unclassified."

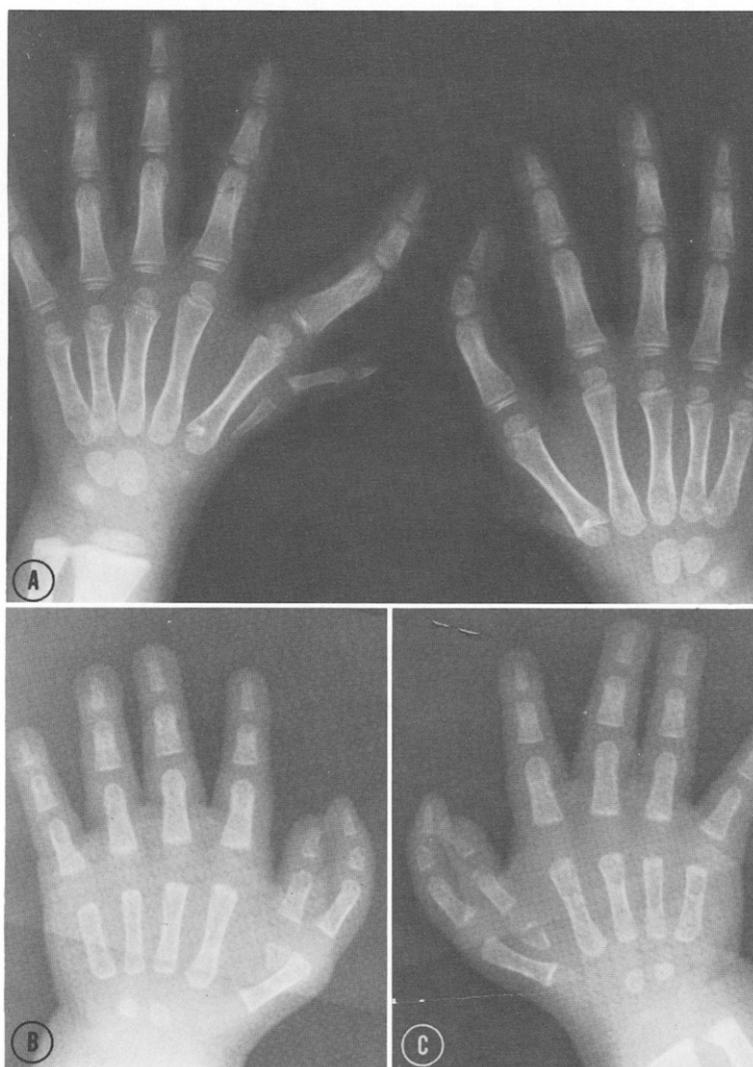
### Treatment

Polydactyly should be treated early. Most operations should be completed by the time the patient is 5 years

of age. The earlier they are done, the more quickly children learn to use their hands correctly.

A number of surgical procedures were required for each patient. Twenty-one patients had a total of 78 operations, for an average of almost four operations per person. One patient with bilateral involvement had five operations, while another patient with bilateral involvement had a total of six operations.

Most operations were for the removal of an accessory digit but the treatment of polydactyly often is not as simple as it appears. It is important to know which thumb the patient is using. For a thumb to function properly, extra structures may have to be transferred from the accessory digit or from another part of the hand. The collateral ligaments should be reconstructed. The joint capsule must be reefed, and K-wire fixation is very important to protect against stretching during heal-



**Fig. 4.** **A**, Roentgenogram of the left hand demonstrates the most common presentation of type C pattern polydactyly as associated with triphalangism. **B** and **C**, Roentgenogram to show bilateral type A pattern polydactyly associated with triphalangism. (Fig. 4, B was published previously and used for another purpose. From Wassel HD: The results of surgery for polydactyly of the thumb. *Clin Orthop* 64:175, 1969.)

ing of the tissues, thereby allowing some joint stability to develop. If an epiphysis is destroyed, shortening or radial or ulnar deviation of the thumb may occur.

In removing an accessory digit, never leave the scar in a straight line. As the thumb grows the scar does not stretch and the part distal to the scar deviates. All straight line scars should be broken with a Z-plasty.

A summary of the surgical procedures is shown in Table II. Fourteen separate ligament reconstructions or fusions were performed. These were done mainly for persistent lateral deviation or instability of the joints following removal of the extra digit (Fig. 6).

In young children the treatment of the triphalangeal thumb with a delta phalanx consists of removal of the delta phalanx and K-wire fixation for at least 6 weeks to prevent displacement of the joint. In adults the problem of the delta phalanx is more complicated. If function is impaired by length or deviation, one should remove the accessory phalanx and most often fuse the joint for stability.

In a type IV polydactyly, the duplicated distal phalanx diverges from the longitudinal axis. Early excision is especially important since the longer it is delayed, the more the supernumerary component tends to

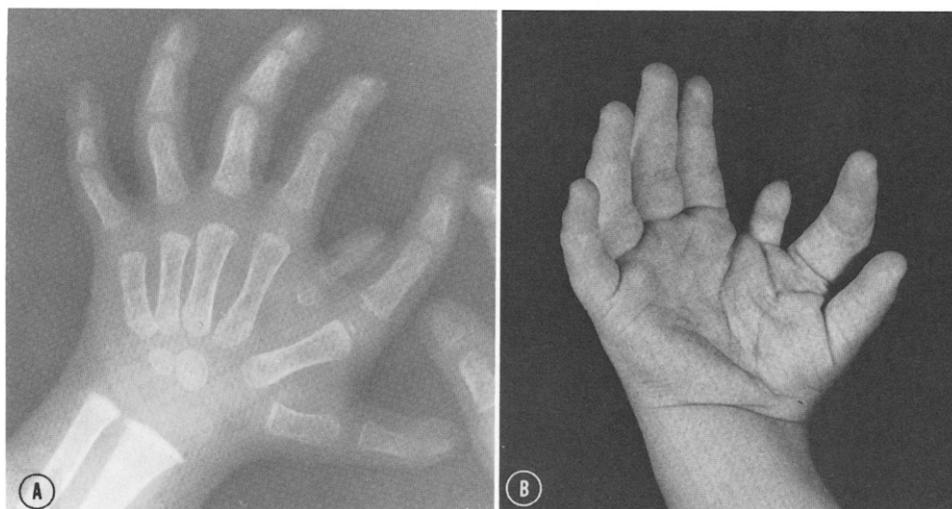


Fig. 5. Roentgenogram (A) and illustration (B) demonstrates a rare anomaly with three thumbs present.

Table II. Surgical procedures

	No.
Removal of polydactyly	30
Reremoval of polydactyly	3
Thumb web deepening	13
Remove delta bone	12
Collateral ligament reconstruction	8
Fuse joint	6
Tendon reconstruction	3
Pollicization	3
Total	78

displace the normal component, thus increasing ulnar or radial deviation. Because of the continued divergence, reconstruction of the ligaments and an osteotomy of the phalanx may be necessary or fusion of the joint may be necessary to avoid further complications. A typical example of deviation of the thumb is shown in Fig. 6, B and C.

If both thumbs are equal in size, appearance, and function, it is best to remove the radially placed digit, because the ulnar collateral ligament thus would remain intact on the ulnar digit. Seldom is a reconstructed ligament as strong as the normal one.

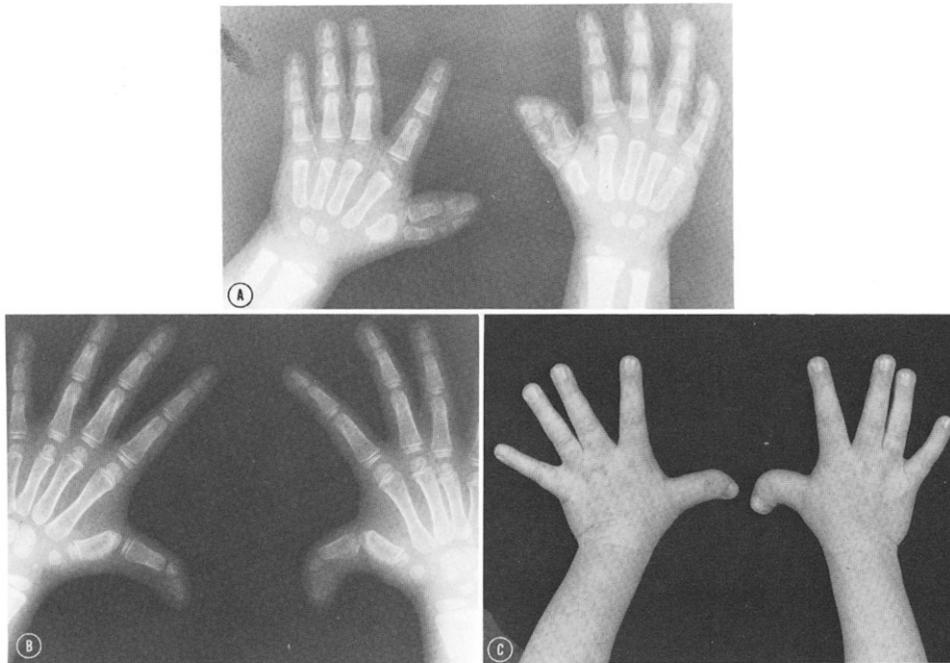
It may be necessary to transfer the distal portion of one thumb onto the more proximal portion of the other. Flexor and extensor tendons may have to be transferred from the duplicated digit so that the final composite will yield the best possible function. When one of two adjoining digits has greater flexor power and the other excels in extension, the latter is amputated and the extensor tendon is transferred to the former. The intrinsic

thenar muscles, such as the abductor pollicis brevis and adductor pollicis, always should be transferred from the amputated digit into the remaining thumb for power and movement.

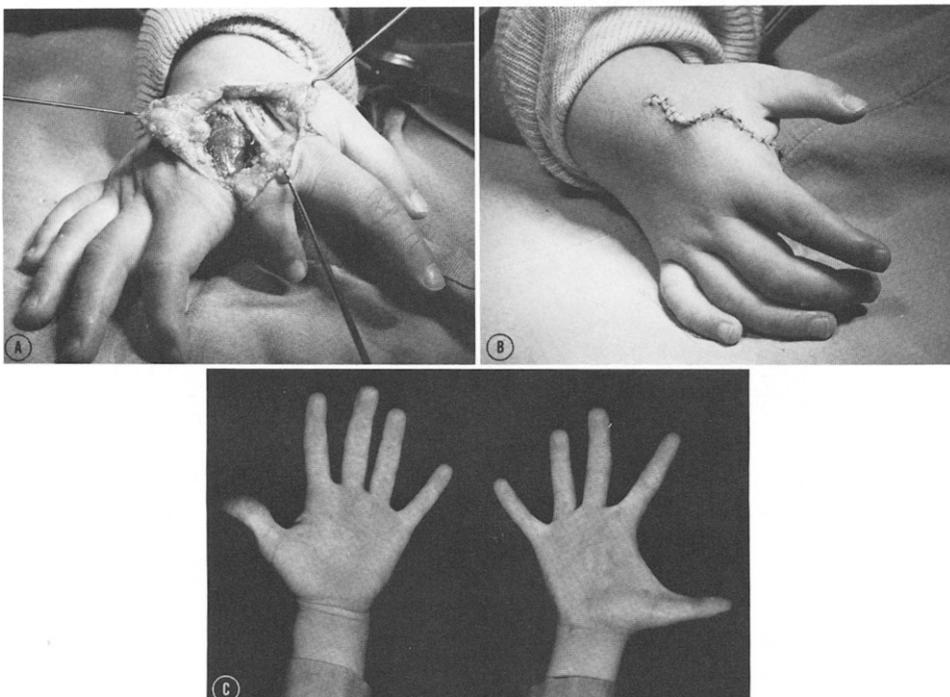
In treating type VII polydactyly, it appears that the best result may be obtained by removing the triphalangeal digit rather than the biphalangeal thumb, even though the latter may be the more rudimentary (Figs. 5 and 7). A more acceptable appearance sometimes may result if the rudimentary thumb is transposed from the ulnar to the radial position and the metacarpal head impaled on the distal end of the metacarpal of the radial triphalangeal thumb as was done in the patient as shown in Figs. 3 and 8. Thus a shorter thumb is made which develops with growth to almost the correct functional length. Sometimes the best surgical result may be obtained by doing a pollicization, by transferring the triphalangeal thumb with rotating flaps, or by a metacarpal osteotomy, as the patient in Figs. 3 and 8 demonstrates.

The problem of the narrow thumb web needs more emphasis on surgical care than has been given to it previously. Twenty-three of the 32 thumbs had very narrow thumb web spaces. Some of the spaces remained narrow even after surgical attempts to deepen the thumb web because of a lack of skin and excessive scarring (Figs. 9 and 10).

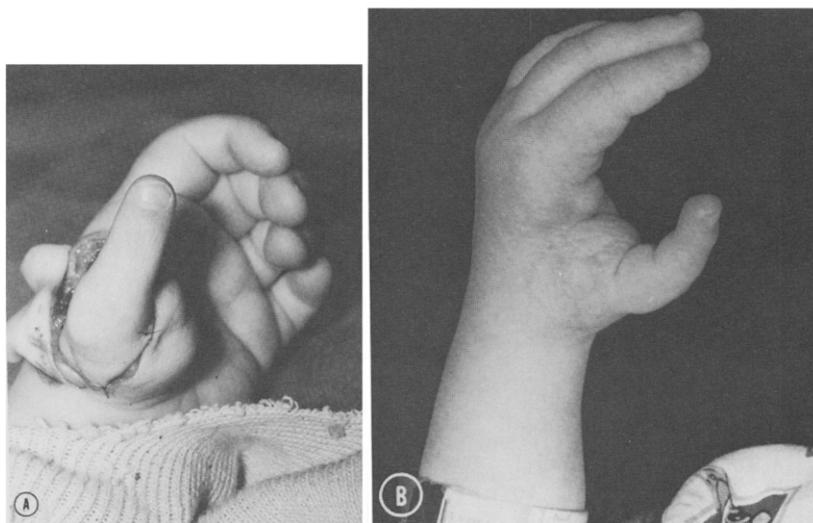
A narrow web space is a major detriment to the functional ability of these children because they lose dexterity and cannot close around large objects with one hand. This then forces these children to become two handed in the majority of their prehensile ac-



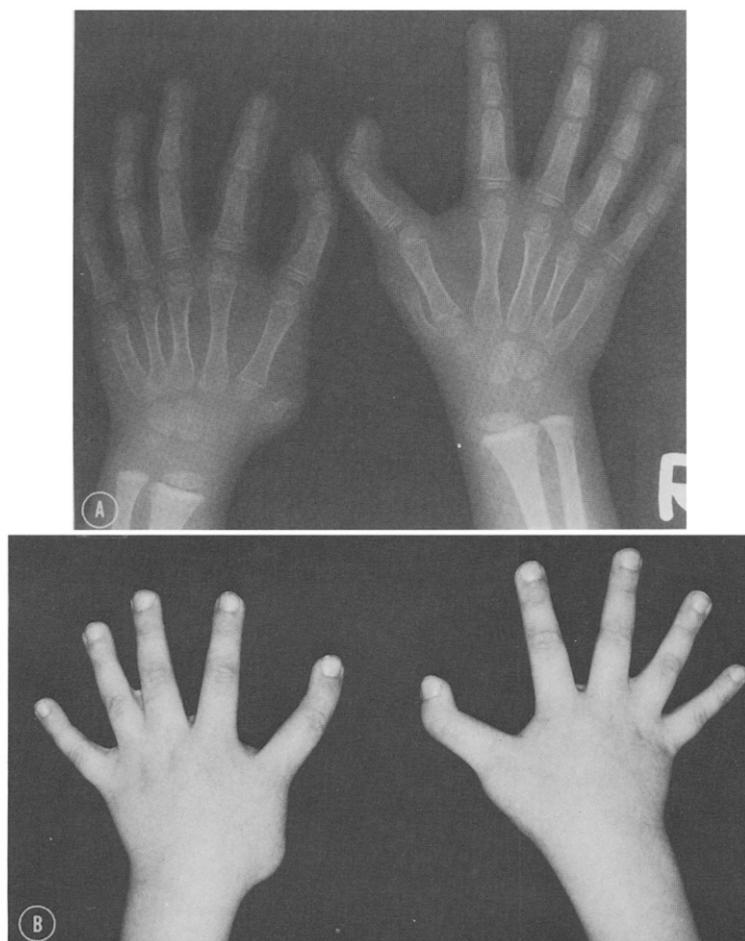
**Fig. 6.** After removal of a type IV polydactyly, the distal phalanx of the thumb fell into a severe, disabling radial deviation. (Fig. 6, **A** and **B**) were previously published and used for another purpose. (From Wassel HD: The results of surgery for polydactyly of the thumb. *Clin Orthop* 64:175, 1969.)



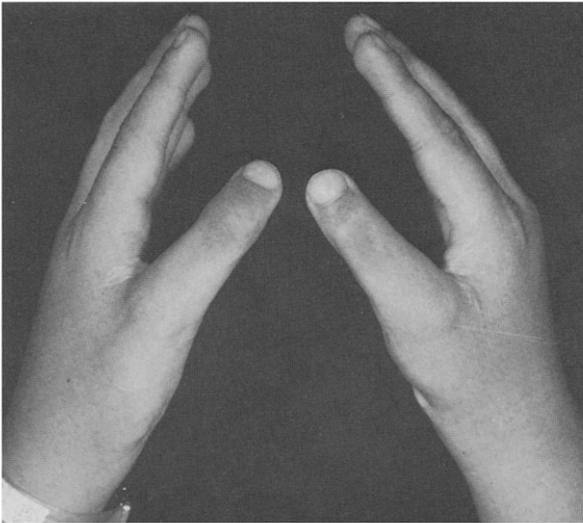
**Fig. 7.** **A**, The patient shown in Fig. 5 had the two ulnarly placed thumbs removed at operation. **B**, The hand following operation. **C**, On the right a good functioning hand is shown 10 years later with an enlarged thumb web space.



**Fig. 8.** **A**, Pollicization and metacarpal rotation of a triphalangeal thumb at the time of operation. **B**, One year later the thumb sits in wide abduction with an adequate web space. (From Wood VE: Treatment of the triphalangeal thumb. *Clin Orthop* 120:188-200, 1976.)



**Fig. 9.** When the patient was 3 weeks of age, both extra thumbs were incompletely removed. At the time of these views (5 years of age) the parents were unhappy with the appearance of both hands.



**Fig. 10.** This same patient as in Fig. 9 is shown 2½ years after removal of the accessory metacarpal and deepening of the thumb web. Even after web plasty, the thumb webs still remain narrow and small.

tivities. We have found that the most useful procedure to create an adequate web space is the four-flap Z-plasty, after the method described by Woolf and Broadbent.<sup>30</sup>

Because there often is not enough skin around the first metacarpal bone, many times the thumb cannot abduct enough when placed in its new position. The lack of skin can be handled very successfully by using a large sliding skin flap from the dorsum of the hand.<sup>31</sup>

One patient showed an enlarged web space. In this case the web space became abnormally wide as a result of surgery (Fig. 7, C). Other than some weakness of pinch, a wide thumb web space was not a functional handicap.

The following outline for the surgical care of polydactyly in association with the triphalangeal thumb is proposed.

### Summary of treatment

- I. General
  - A. Remove polydactyly
  - B. Remove *all* accessory parts
  - C. Remove delta bone
- II. Type IV polydactyly
  - A. Treat *very* early—6 months
  - B. Equal digits—remove radial one
  - C. Reconstruct ligaments—K-wire fix for long periods
  - D. Unstable joint—fuse

- E. Deviation phalanx—osteotomy
  - F. Transfer portions of digits for best composite
- III. Type VII polydactyly
    - A. Remove triphalangeal thumb even if biphalangeal more rudimentary
    - B. Pollicize with large dorsal flap
    - C. Metacarpal osteotomy
  - IV. Thumb web contracture
    - A. Four-flap Z-plasty (Woolf)
    - B. Large dorsal rotation flap

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### REFERENCES

1. Demarinis F, Wildervanck LS: Pre-axial polydactyilia (double thumb and triphalangia.) *Ned Tijdschr Geneesk* 104:2169, 1960
2. Dubreuil-Chambardel L: Un case hyperphalangie du pouce. *Gax Centre Tours* 15:25, 1910
3. Hilgereiner H: Ueber Hyperphalangie des Daumens. *Beitr Klin Chir* 54:585, 1907
4. Barsky AJ: Congenital anomalies of the thumb. *Clin Ortho* 15:96, 1959
5. Mueller W: Contributions to knowledge of the triphalangeal thumb. *Arch Klin Chir* 185:377, 1936
6. Nylander ES: Pre-axiale Polydaktylie in Funf Generationen Einer Schwedischen Sippe Upsala Lak. *Foren Forh NF* 36:275, 1931
7. Wood VE: Treatment of the triphalangeal thumb. *Clin Ortho* 120:188, 1976
8. Maurizio R: Primus digitus varus, triphalangism of the thumb and radial polydactyilia. *Arch Putti Chir Organi Mov* 19:449, 1964
9. Milch J: Triphalangeal thumb *J Bone Joint Surg* 33-A:692, 1951
10. Polinelli U: A case of familial hyperphalangia of the thumbs. *Minerva Nipiol* 12:373, 1962
11. Townes PL, Brocks ER: Hereditary syndrome of imperforate anus with hand, foot, and ear anomalies. *J Pediatr* 81:321, 1972
12. Komai T, Ozaki Y, Inokuma W: A Japanese kindred of hyperphalangism of thumbs and duplication of thumbs and big toes. *Folia Hered Pathol* 2:307, 1953
13. Swanson AB, Brown KS: Hereditary triphalangeal thumb. *J Hered* 53:259, 1962
14. Velichkon LG: Problem of the biphalangy or triphalangy of the first digit. *Arkh Anat Gistol Embriol* 58:87, 1970
15. Windle BCA: The occurrence of an additional phalanx in the human pollex. *J Anat.* 26:100, 1891
16. Lapidus PW, Guidotti FP: Triphalangeal bifid thumb. *Arch Surg* 49:228, 1944

17. Lapidus PW, Guidotti FP, Coletti CJ: Triphalangeal thumb—report of six cases. *Surg Gynecol Obstet* 77:178, 1943
18. McMurtry R: Personal communication, Sunnybrook Medical Center, University of Toronto, 1973
19. Bienvenue F: Un Case de Pouces Supplementaire a Trois Phalanges. *Rev Orthop Chir Par* 111:91, 1912
20. Francesconi G: Clinical considerations on a thumb malformation of a familial nature. *Minerva Chir* 21:618, 1966
21. Hass SL: Three-phalangeal thumb. *Am J Roentgenol Radium Ther Nacl Med* 42:677, 1939
22. Jager M, Refior HJ: The congenital triangular deformity of the tubular bones of the hand and foot. *Clin Orthop* 81:139, 1971
23. Kirmission E: Pouces a Trois Phalanges. *Rev Orthop Par* 10:256, 1956
24. Kristjansen A: Supernumerary phalanx in the thumbs. *Hyperphalangeal pollicis*. *Hospitaltid* 69:109, 1926
25. Millesi H: Deformations of the fingers following operations for polydactyly. *Klin Med (Vienna)* 22:266, 1967
26. Wiedemann HR: Hinweis auf eine derzeitig Haufung hypo- and aplastischer Fehlbildungen der Gliedmassen. *Med Welt* p. 1863, 1961
27. Wassel HD: The results of surgery for polydactyly of the thumb. *Clin Orthop* 64:175, 1969
28. Miura T: Triphalangeal thumb. *Plast Reconstr Surg* 58:587, 1976
29. Tuch BA, Lipp EB, Larsen IJ, et al: A review of supernumerary thumb and its surgical management. *Clin Orthop* 125:161-162, 1977
30. Woolf RM, Broadbent TR: The four-flap Z-plasty. *Plast Reconstr Surg* 49:48, 1972
31. Tajima T, Watanabe Y, Uchiyama J: Treatment and study of the hypoplastic thumb. *Jpn J Plast Reconstr Surg* 10:227, 1967

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