主　論　文



Original

# Histological Analysis of Triphalangism Associated with Polydactyly of The Thumb

（母指多指に合併した三指節母指の組織学的分析）

Hitomi MAKINO, Takayuki MIURA,
Ryogo NAKAMURA and Hiroshi NOGAMI

（牧野仁美、三浦隆行、中村蓼吾、野上宏）

ABSTRACT Wassel's classification of thumb polydactyly has some problems especially in Ⅳ and Ⅶ categories because it depends solely on X-ray findings. In order to improve his thumb polydactyly classification, we histologically examined untreated 182 bifid thumbs from 171 patients at Branch Hospital, Nagoya University in the past 8 years. Ninety-five of these cases had proximal phalangeal separate type of polydactyly corresponding to Wassel's Ⅳ and Ⅶ categories. Forty-five thumbs of these categories were excised from 44 patients and classified into type Ⅰ (24), type Ⅱ (12), type Ⅲ (4), and type Ⅳ (5) in accordance with Kanno's classification. In distal phalanges, normal cartilage development was observed in Kanno's type Ⅰ, whereas proximally elongated cartilaginous tissue was formed in the type Ⅱ. Three distal phalanges from types Ⅱ and Ⅲ showed depression at the ulnar side of elongated cartilage of the distal phalanx where immature chondrocytes transversely distributed. Three cases with incomplete and one case with complete joint formation were formed in elongated cartilage of the distal phalanx, and endochondral ossification developed in the central area of elongated cartilage of the distal phalanx in 4 cases from types Ⅲ and Ⅳ. All of the cases showed only minimal mobility of their involved joints. Their tendon development did not correspond to their anatomical characteristics. Result of histological studies indicated that there was no distinct difference between Wassel's category Ⅳ and category Ⅶ. It was postulated that the triphalangism belonging to Wassel's category Ⅶ should

be classified according to its type of bifurcation rather than its number of phalanges.

Key words : thumb polydactyly, triphalangism, Wassel's classification, symphalangism, histology, anatomy

## INTRODUCTION

Triphalangism is not uncommon in cases of thumb polydactyly bifurcated at the level of metacarpophalangeal joint, and occurs radially, ulnarly, or bilaterally. The radial type, with duplication of the proximal phalanx, is most commonly seen. Wassel (1969) categorized thumb polydactyly into 7 types (Fig.1). He grouped 2 phalangeal type into 6 categories according to their bifurcation types of phalangeal development, and especially indexed triphalangeal thumb into Type Ⅶ. This classification is, however, not exactly correct because type change from 2 phalangeal type into 3 phalangeal type is not uncommon in fast growing infants. Kanno et. al.(1978) classified thumb polydactyly of the proximal phalangeal separate type into 4 categories: 2 segments (type Ⅰ); wide space between the proximal and distal phalanges (type Ⅱ); 3 segments containing epiphyseal bone(type Ⅲ); and those containing phalangeal bone(type Ⅳ) (Fig.2).He further examined excised thumbs and reported that type Ⅱ contained uniform hyaline cartilage in the space between the distal and the proximal phalanges. One case of type Ⅱ, however, displayed ossification at the center of its cartilage 4 months after the first examination and the category was changed to type Ⅲ. Types Ⅲ and Ⅳ included 2 types respectively: one with only one interphalangeal(IP) joint, and the other with 2 IP joints, he described. We studied the histological and anatomical findings of thumb polydactyly belonging to Wassel's Ⅳ and Ⅶ categories with the object of improving Wassel's classification. Special

care was taken to define tendon attachment in triphalangism.

## MATERIALS AND METHODS

One hundred and seventy-one untreated patients with 182 bifid thumbs were seen at Branch Hospital, Nagoya University from 1984 to 1991. They were categorized as follws in accordance with Wassel's classification: I (8), II (26), III (13), IV (50), V (22), VI (18), and VII (45). There were 95 cases of proximal phalangeal separate type belonging to Wassel's IV and VII. Forty-five thumbs were excised from 44 patients. There were 32 right thumbs and 13 left. All were triphalangism of the radial type, and were further categorized as type I (24), type II (12), type III (4), and type IV (5) in accordance with Kanno's classification. Patients' ages at surgical operation varied from 4 to 11 months with an average of 6.6 months. Digital tendons were examined microscopically during surgery. Excised phalanges were then sectioned longitudinally at the plane parallel to the nail in order to observe their interior structures. They were then fixed in 20% formalin solution, and were later decalcified, dehydrated, and embedded in paraffin. Serial sections were made and stained with hematoxylin and eosin. Alcian blue and neutral red staining was also done.

## RESULTS

Histological examination of 45 cases of proximal phalangeal separate thumb polydactyly revealed that epiphyseal cartilage development was 22.2% to 44.4% with an average of 31.7% of the length of the distal phalanx. It was in accordance with

Kanno's type I (Fig. 3A). In Kanno's type II, epiphyseal cartilage elongated and tapered toward the ulnar side at the space between the distal and proximal phalanges. There were hypertrophic chondrocytes which seemed to be a initial sign of epiphyseal ossification in 9 cases of type II (Fig. 3B), and their elongated cartilage varied from 43.5% to 75.0% with an average of 59.2%. These rates were higher than those of type I ($p < 0.001$), but some of them were difficult to classify into type I or type II categories because they were made up of various amount of epiphyseal cartilage. There were slight depression at the mid-level of the elongated cartilage at the ulnar side in 2 cases of type II and one of type III. There were relatively small chondrocytes at the area of above mentioned depressions (Fig. 3C). These chondrocytes were similar to those seen under the articular surface. One case of type III and 2 cases of type IV had incompletelyd eveloped joints between the distal phalangeal bone and the interventional bone. The interventional bone was partially connected cartilaginously with distal phalanx on the radial side, and on opposite side, there was a crevice covered with articular cartilage (Fig. 3D). One case of type IV had normal joint development with joint capsule (Fig. 3E), but they were not seen in those cases with crevice formation. The joints displayed little or no mobility. Four cases of types III and IV only displayed ossification at the elongated epiphyseal cartilage of their distal phalanges.

The features of the tendon are summarized in Table I. Two

thumbs out of 15 with both extensor and flexor tendons showed chiasma structures similar to flexor digitorum superficialis. The tendon development in cases of types Ⅱ, Ⅲ, and Ⅳ, was better than in cases of type Ⅰ. It seemed, however, that the tendon structures depended on the development of polydactylous components rather than the characteristics classified by Kanno. Flexor tendons were attached to the elongated epiphyseal cartilage of the distal phalanges or the interventional bones as if they were flexor digitorum superficialis in cases of types Ⅱ, Ⅲ, and Ⅳ, but the tendon in one case of type Ⅳ was inserted into the distal phalanx over the interventional bone as if it was a flexor digitorum profundus.

Relationship between patient age and each type varied as follows: 6.7 months(type Ⅰ), 6.7months(type Ⅱ), 6 months(type Ⅲ), and 6.5 months(type Ⅳ).

## DISCUSSION

Ogino et al.(1983) reported histological findings of interventional bone in triphalangism associated with polydactyly of the thumb. They concluded that Kanno's type Ⅲ was essentially the same as Kanno's type Ⅱ, and only difference was their maturation stage. They also noted that Kanno's type Ⅳ had normal joint development between each phalanx. Islam and Fujita (1991) considered that the elongated epiphyseal cartilage of the distal phalanx or the interventional bone corresponds to the middle phalanx. They classified it into three types: (A) having no articular formation, (B) with joint formation between each

phalanx, and (C) with joint formation only between the middle and the proximal phalanges. They also suggested that the thumb originally had three phalanges, but it evolved with two phalanges as a result of the fusion of the distal and middle phalanges or the obliteration of the middle phalanx. It is very interesting to note that one of the results of our study showed agreement with Islam and Fujita's(1991) supposition. That is, triphalangism associated with polydactyly of the thumb once were normally jointed between the distal and middle phalanges and gradually fused together on the radial side. They evolved to one segment with epiphyseal cartilage ossification at the elongated epiphyseal cartilage of the distal phalanges. The elongated cartilage gradually shortened and finally became what Wassel classified as type Ⅳ. We saw no cases which fell into Islam's classification A, but there were 2 cases of the rudimentary type which did not have articular formation. Takahara et al.(1989) reported their experimental model of symphalangism of the distal interphalangeal joint (DIPJ) associated with hypoplasia of the middle and the distal phalanges. They(1990) went on to say that this condition occurred as a consequence of excessive mesenchymal cell death, and that aplasia of the DIPJ appeared in accordance with distal defects in tendons which are normally inserted on the DIPJ(1991). Their conclusion supports our hypothesis that the distal phalanx fused with the middle phalanx and gradually evolved into one segment. And our cases of types Ⅱ, Ⅲ, and Ⅳ, with the exception of one case of type Ⅳ,

displayed distal defects in tendons such as Takahara et al.(1991) described. We concluded that Wassel's category Ⅳ was not clearly distinguished from category Ⅶ. It is our opinion that the triphalangism belonging to Wassel's category Ⅶ should be classified according to its type of bifurcation rather than its number of phalanges. Our study didn't reveal any corresponding similarities between the age and the stage of ossification. We supposed, however, that Kanno's typeⅡ would have eventually matured to typeⅢ or type Ⅳ if we allowed this condition to progress without any treatment. We also expect that both age and development of thumb polydactyly component may have relation to the stage of ossification.

Nogami(1984) mentioned that the critical period in utero of thumb polydactyly development was same as that of the thumb triphalangism based on his experimental works. He also concluded that these defects had common genesis, and their difference might be due to the difference of amount and site of lesions occurred during mesenchymal condensation. He went on to say that the extra phalanx was identical to the middle phalanx because chondrification of the extra phalanx in thumb triphalangism coincided with the middle phalanges of other fingers. Seo et al. (1977) suggested that thumb triphalangism is the outcome of additional segmented degeneration and/or fusion in thumb polydactyly. Miura(1976) reported that the wedge shaped interventional bone in thumb triphalangism often appeared on the radial side, while radial type triphalangeal deformities of

thumb polydactyly appeared more often than did the other types. He also postulated that the occurrance of thumb triphalangism was due to the failure to separate of bifid thumb. Our opinion is that thumb triphalangism is not simply an atavism, but dysplasia of the extra segment in triphalangeal thumb polydactyly. We believe that the radial type of triphalangeal thumb polydactyly corresponds to thumb triphalangism with wedge shaped bone formation on the radial side, because the incidence of these cases are the highest in each group. The ulnar type corresponds to wedge shaped bone thumb triphalangism on the ulnar side, and the bilateral type corresponds to thumb triphalangism having interventional middle phalanx bone formation (Fig.4). We further hypothesize that they may also have some common characteristics such as: their incidence being fewer in number and more dysfunctional than the radial type. The lower incidence of thumb triphalangism as compared to triphalangeal thumb polydactyly may be the result of the infrequency in arising of bifid thumbs' dysplasia.

ACKNOWLEDGMENT

We are sincerely indebted to Dr. Suzuki, Tokai Hospital, for his valuabule assistance and advices.

REFERENCES

Islam, S. and Fujita, S. (1991) Triphalangism in thumb polydactyly: An anatomic study on surgically resected thumbs. Plast. Recon-

str. Surg., 88:831-836.

Kanno, T., Yoshizawa, N., Uetake, M., Kamata, M., Nishimura, T. and Tsuchiya, I. (1978) Study of 47 cases of polydactyly of the thumb. Orthp. Surg., 29:1561-1563(Japanese)

Miura, T. (1976) Triphalangeal thumb. Plast. Reconstr. Surg., 58:587-594

Nogami, H. (1984) Formation mechanism of congenital anomalies in the hand. Seikeigeka Mook, 35: Congenital Anomalies of the Hand (Tsuge, K. ed.), Kanehara Shuppan, Tokyo, 28-40(Japanese)

Ogino, T., Ishii, S., Usui, M., Minami, A., Fukuda, K., Minami, M. and Kato, S. (1983) Histologic feature of the extra phalanx acommpanied with triphalangeal thumb polydactyly. Orthop. Surg., 34:1789-1791(Japanese)

Seo, Y., Watari, S., Yamamoto, S. and Tsuge, K. (1977) Experimental thumb deformity and clinical cases. Plast. Surg., 20:387-388 (Japanese)

Takahara, M., Ogino, T., Minami, A., Kato, H. and Ohshio, I. (1989) Experimental study on symphalangism. J. Jpn. Soc. Surg. Hand., 6:798-802(Japanese)

Takahara, M., Minami, A., Kato, H., Ohshio, I. and Ogino, T. (1990) Experimental study on symphalangism: 2nd report(a study of the process of joint fusion). J. Jpn. Soc. Surg. Hand., 7:783-787 (Japanese)

Takahara, M., Minami, A., Itoga, H., Ohshio, I. and Ogino, T. (1991) Experimental study on symphalangism: 3rd report(development of tendon). J. Jpn. Soc. Syrg. Hand., 8:489-493(Japanese)

Wassel, H. D. (1969) The results of surgery for polydactyly of the thumb. Clin. Orthop., 64:175-193

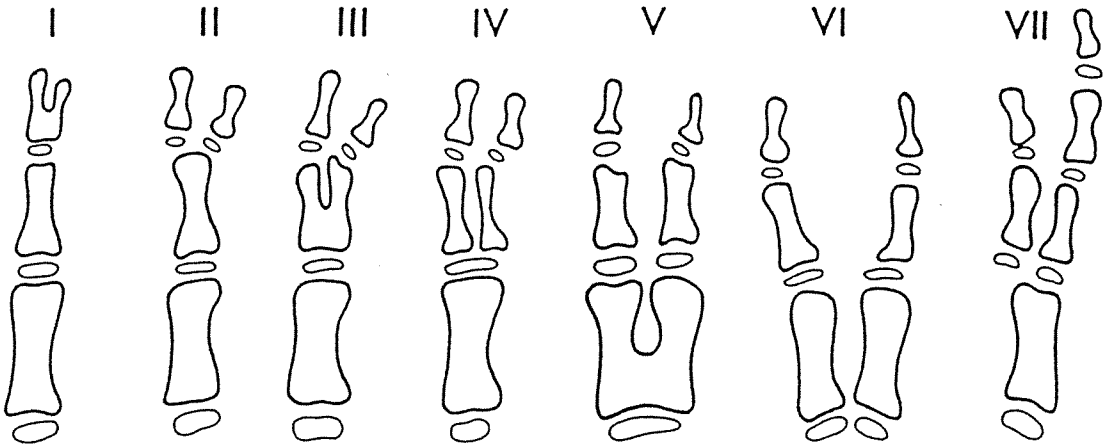



Fig. 1 Wassel's classification of thumb polydactyly.

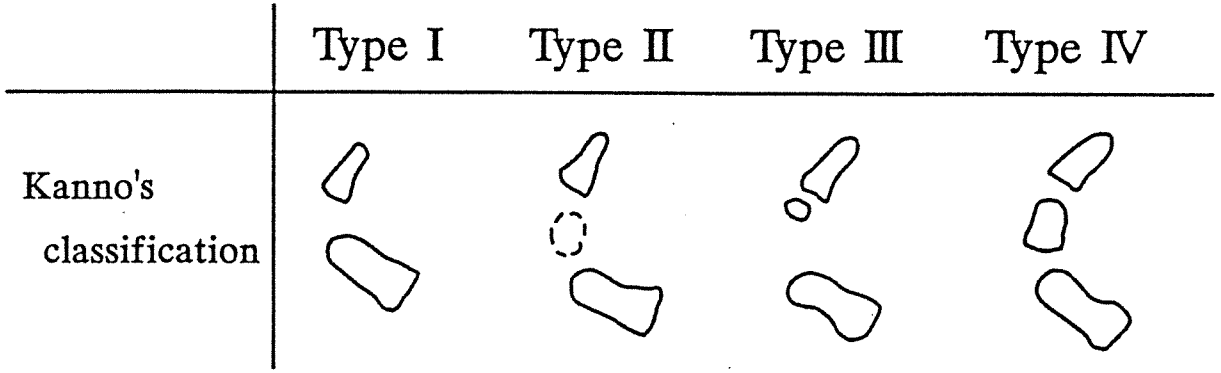



Fig. 2

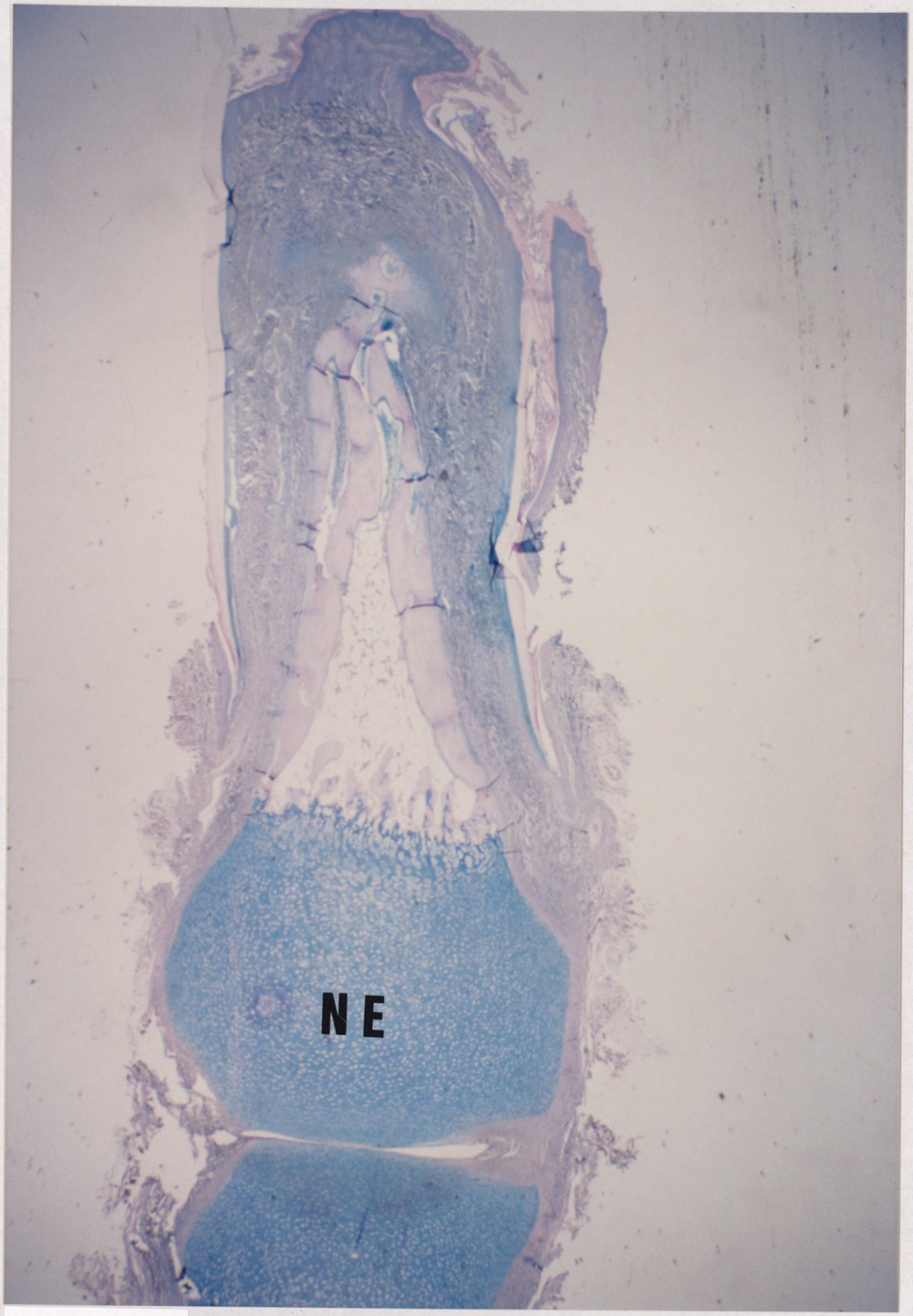



Fig. 3A

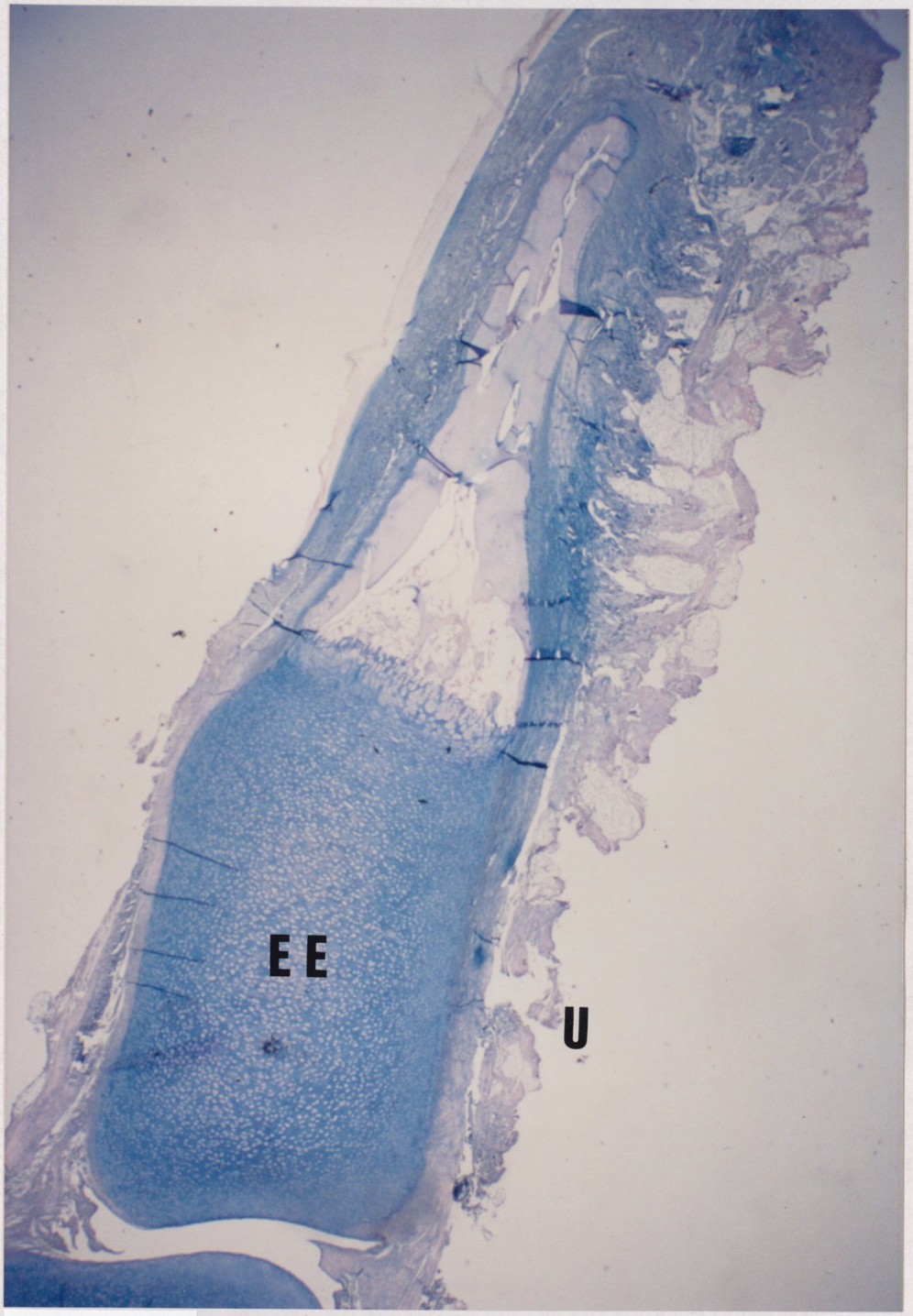



Fig. 3B

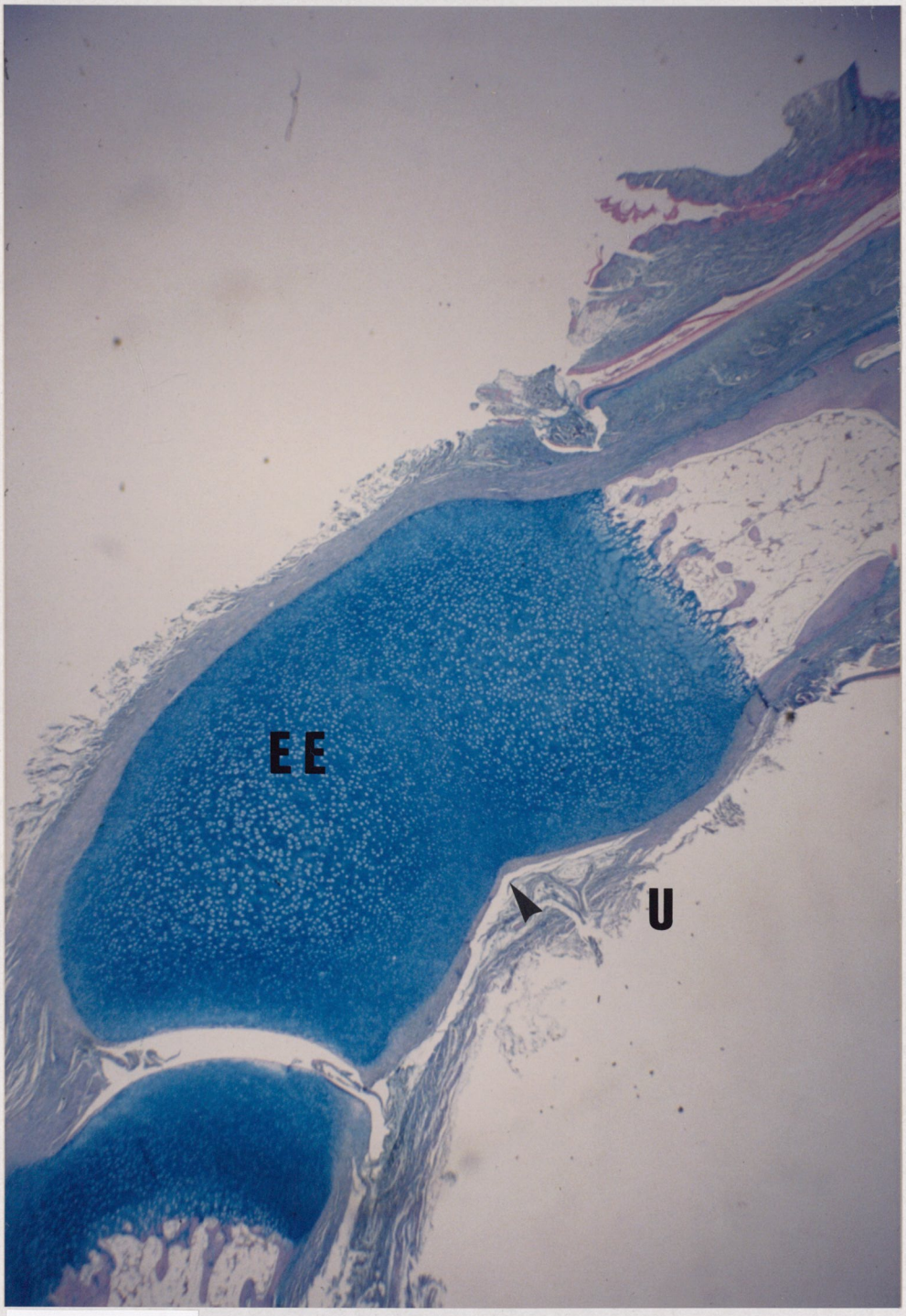



Fig. 3C

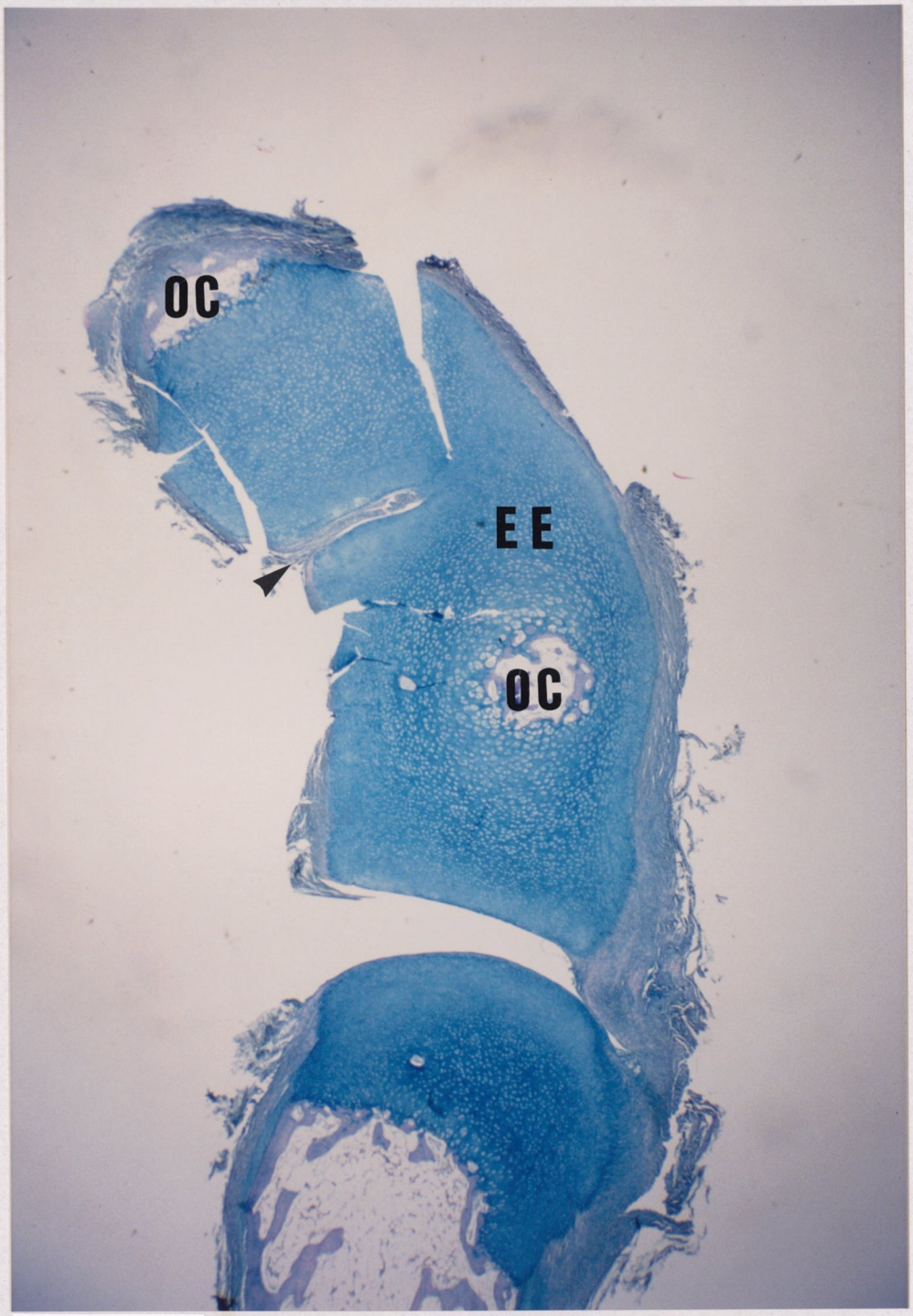



Fig. 3D

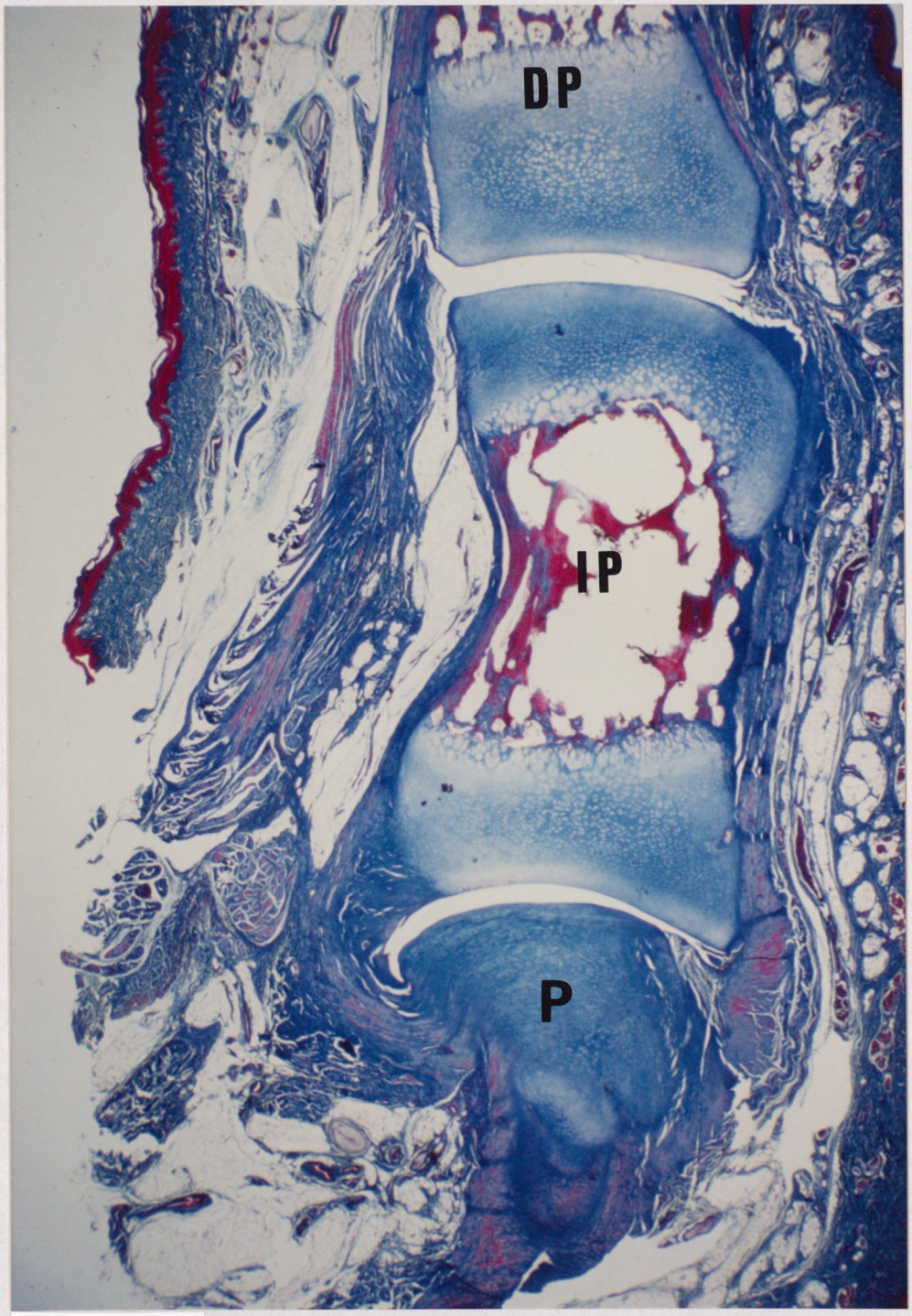
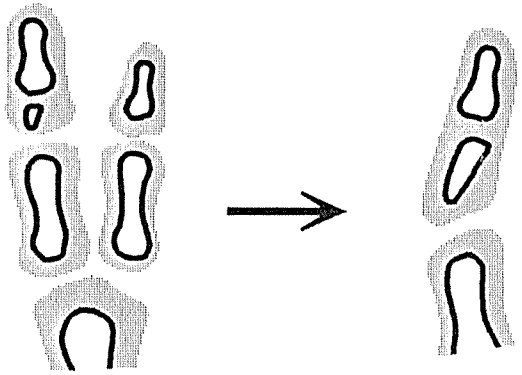
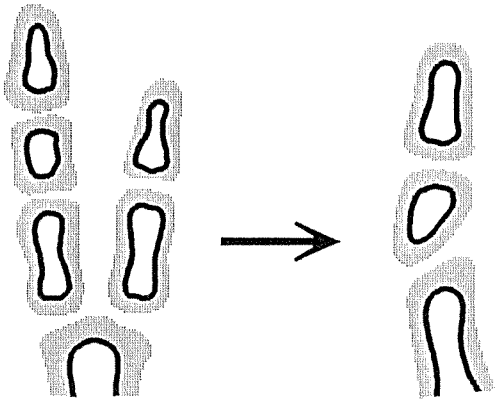


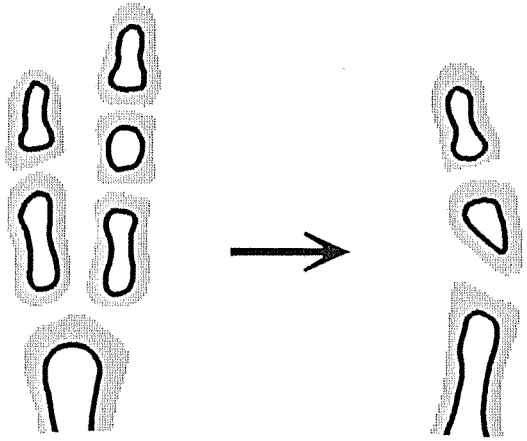

Fig. 3E



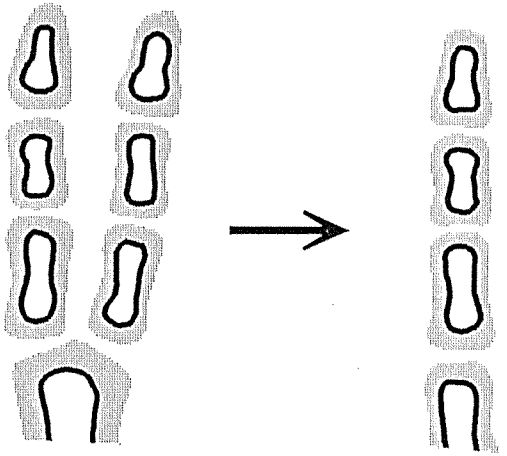
A



B



C



D

Fig. 4

## FIGURE LEGENDS

Fig. 1 Wassel's Classification of thumb polydactyly

I Bifid distal phalanx

II Duplicated distal phalanx

III Bifid proximal phalanx

IV Duplicated proximal phalanx

V Bifid metacarpal

VI Duplicated metacarpal

VII Triphalangism

Fig. 2 Kanno's classification of thumb polydactyly of proximal phalangeal separate type

Fig. 3 Photomicrographs showing longitudinal sections of excised polydactyly of the thumb

A Distal phalanx with normal epiphyseal cartilage(NE). (Kanno's type I : 6.5 months old, × 40)

B Distal phalanx with proximally elongated epiphyseal cartilage(EE), tapering toward ulnar side(U). (Kanno's type II : 6 months old, × 40)

C Distal phalanx with proximally elongated epiphyseal cartilage(EE), showing a slight depression on the ulnar side(U). Note : hypertrophic chondrocytes are divided into 2 groups by immature resting chondrocytes(arrow). (Kanno's type II : 6 months old, × 40)

D Distal phalanx with proximally elongated epiphyseal cartilage(EE), showing crevice formed with articular cartilage and filled with soft tissues similar to joint capsule

(arrow) Note : two ossification centers(OC) bordered by the crevices. (Kanno's type Ⅳ : 6 months old, ×40)

E A normal joint development between distal phalanx(DP) and interventional phalangeal bone(IP). P:proximal phalanx. (Kanno's type Ⅳ : 8 months old, ×40)

Fig.4 Hypothetical correspondence of triphalangeal thumb polydactyly with thumb triphalangism

A Radial triphalangeal type (with epiphyseal ossification)

B Radial triphalangeal type (with phalangeal ossification)

C Ulnar triphalangeal type

D Bilateral triphalangeal type

Table 1 Tendon development corresponding to Kanno's classification

| Kanno's classification | type I | type II | type III | type IV |
|---|---|---|---|---|
| No tendons | 8 | 2 | | 1 |
| Extensors only | 5 | 2 | | |
| Flexors only | 2 | 1 | 2 | |
| Both extensors and flexors | 5 | 6 | 2 | 2 |
| Unknown | 4 | 1 | | 2 |
| Total | 24 | 12 | 4 | 5 |