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TITLE

Use of the duplication range concept for understanding morphology and predicting prognosis in thumb polydactyly

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congenital abnormalities; deformity; polydactyly; thumb

ABSTRACT

This study systematically and comprehensively analysed 129 thumb polydactylies in 122 patients using a duplicating range concept based on the level of skin and skeletal bifurcation. Numerical levels were defined along the longitudinal axis of the ulnar thumb duplicate from distal to proximal: level 0 (thumb tip) to level 6 (carpometacarpal joint). The relationships between duplication range and morphological parameters were evaluated. Nail asymmetry was associated with skin bifurcation levels 0 to 2. Proximal phalangeal asymmetry and interphalangeal joint deviation, associated with skin bifurcation levels were assigned scores of 1 to 2.5 and 1 to 2, respectively. Metacarpophalangeal joint deviation had a bimodal distribution, at levels 1.5 and 4 of the longitudinal axes. Morphological similarity was found in patients with the same duplication range. The duplication range concept could potentially improve our

understanding of morphology variation in thumb polydactyly.

Level of evidence: III

INTRODUCTION

The most widely used categorization system for thumb polydactyly is the Wassel classification (Wassel, 1969) based on the skeletal bifurcation level and the presence of triphalangism. Despite its popularity, the classification has been criticized for failing to identify individual morphological features, even among individuals with the same skeletal bifurcation level. Recently, new classification systems have been proposed; Chung et al., (2013) described a system according to the osseous continuity between the two thumbs while the Rotterdam classification (Zuidam et al., 2008) is based on the combination of the skeletal bifurcation level and several associated abnormalities, including triplication, triphalangism, hypoplasia, deviation, and symphalangism. In the latter, associations are labelled as suffixes and combined with their positional information (radial, ulnar, or both). Recent studies have demonstrated high reproducibility and efficacy of the Rotterdam classification, especially when hypoplasia is added to the categorization based on the skeletal bifurcation level (Hu et al., 2021).

While the Rotterdam classification offers a vast improvement in terms of classifying all different thumb polydactyly phenotypes, we have found there are still variations in the appearances and skeletal characteristics among patients within the same category. Such variations are not negligible; for example, the asymmetric morphology of the nail and pulp is likely to influence postoperative cosmesis (Goldfarb et al., 2008; Tien et al., 2007). Skeletal morphological abnormalities are prognostic factors for postoperative deformation such as severely deviated joints in Wassel type IV thumb polydactyly (Hong et al., 2021; Kim et al., 2017; Lee et al., 2013; Luangjarmekorn et al., 2021) and asymmetric morphology of phalanges (Saito and Morimoto, 2021). The presence of hypoplastic elements is also important; even a well-developed ulnar thumb with a very hypoplastic 'floating' radial thumb has the potential to cause joint deviation (Hanaka et al., 2021; Lourie et al., 1995; Saito et al., 2018). With most of the existing classification systems focused on skeletal morphology, there is a need to revisit the soft tissue abnormalities which may lead to a better understanding of the morphological variations. The syndactylous web between the two thumb duplicates is an area of conglomeration, i.e., soft tissues are shared by the two thumbs proximally whereas they are independent distally, albeit hypoplastic. In this area, bifurcating flexor pollicis longus (FPL) tendons pass through wide pulley structures (He and Nan, 2016; Saito et al., 2021; Tien et al., 2007) and the thenar musculature inserts separately into the radial and ulnar thumbs, causing abduction–adduction imbalances (Saito et al.,2021). We proposed the term "duplication range" to refer to the region between the distal margin of the syndactyly and proximal skeletal bifurcation levels. We hypothesized that the morphological characteristics of thumb polydactyly depend on the contents in this duplication range. The purpose of this study was to elucidate the morphological diversity of thumb polydactyly by systematically and comprehensively assessing the relationship between thumb morphology and the duplication range in a series of patients.

PATIENTS AND METHODS

Study design

This retrospective study was conducted with the approval of the ethics review board of our institution. Written informed consent for the research use of medical records, radiographs, and photographs was obtained from parents of all patients at the time of consultation. An opt-out method allowed patients to decline participation in this study, in accordance with the Declaration of Helsinki. The study included patients with thumb polydactyly who were treated at our institution from January 2010 to December 2021. Patients who did not have radiographs or photographs taken before their initial thumb polydactyly surgery were excluded. The stratification and classification were performed by three hand surgeons (one had level 2 and the others had level 4 experience (Tang and Giddins, 2016), The three hand surgeons were briefed on the duplication range classification using a referential diagram before providing ratings.

Stratification by radiological appearance

Initially, the thumbs were broadly stratified by the radiological appearances using the Wassel classification (Figure 1a). Duplications that had an articulation or osseous connection, and complete osseous components of near-normal morphology, were defined as the non-floating type, and these were further classified into evenly and ulnar-dominant types. In addition, thumb polydactylies with a non-articulated connection on the radial side was defined as the floating or rudimentary type (Figure 1b). The floating type was further divided into three subgroups according to the number of osseous components contained in the radial thumb: monophalangeal, biphalangeal, and triphalangeal types. Finally, a ‘rudimentary type’ was also included, to describe the presence of a nubbin-like radial thumb as divided into two subgroups: thumbs with a thick base were classified into the sessile type, and those with a narrow stem were classified into the pedunculated type. Thumb duplications with a triphalangeal ulnar thumb were categorized independently of the non-floating subtypes.

Sub-classification of structures in the duplication range

As mentioned, the duplication range was defined as the area between the skin syndactyly (distal) and skeletal (proximal) bifurcation levels (Figure 2). Within this range, the normally unilateral soft tissues are duplicated and the arrangement of these soft tissues are dependent on the types of duplications (non-floating or floating). To correspond the soft tissues with the skeletal morphology, the level of skeletal bifurcation was further divided into levels as according to the Wassel classification (Figure 3a). Duplications proximal to the base of the metacarpal, i.e., at the level of the trapezium, were not defined because the majority of patients (98%) in this study were treated before the age of 2 years, before complete ossification of the carpal bones. In order to accurately express variations in location, intermediate levels were defined in addition to the six skeletal bifurcation levels (e.g., level 2.5 corresponds to the level between level 2 and level 3) (Figure 3a). In addition, level 0 represented phenotypes in which the skin was completely fused distally, level 0.5 was defined for phenotypes with a cutaneous notch that reached the nail bed and level 1 corresponded to the level of the nail matrix. In Wassel IV non-floating thumbs, we observed that the level of duplication can vary at the level of the MCP joint. To account for this, a further sub-classification was used: sublevels 4d, 4m, and 4p corresponding to the proximal end of the proximal phalangeal epiphysis, the centre of the joint, and the distal end of the metacarpal head, respectively (Figure 3b).

Morphological analyses

Photographs and radiographs taken at around 1 year of age (before surgery) were used to assess the osseous and nail morphology of the ulnar thumb. Parameters used for morphological analyses included nail asymmetry, proximal phalangeal head asymmetry, proximal phalangeal metaphysis asymmetry, radial deviation of the IP joint, and ulnar deviation of the MCP joint (Figure 4). After thumbs were stratified by the degree of development, the relationship between the duplication range and each morphological parameter was evaluated. For type IV duplications, the detailed relationship between the skin and skeletal bifurcation levels was evaluated but not for the others. The data were expressed as a percent frequency for each duplication range subtype. The Fleiss' Kappa statistic was used to assess the inter-reliability among the three hand surgeons.

RESULTS

Patients

A total of 129 hands (88 right; 41 left) of 122 patients were eligible. The distribution of duplication range subtypes is as shown in Figure 5. Type IV was the most common (41 hands: 32%), followed by Type II (17 hands: 13%) and Type V (10 hands: 8%). 95 had the non-floating type (74%), 25 had the floating type (19%), and nine had the rudimentary type (7%). The 95 thumbs with the non-floating type were further subdivided into 32 with the evenly dominant type, 51 with the ulnar dominant type, and six with a triphalangeal ulnar thumb. Six hands were unclassifiable. In type IV thumbs, those with the ulnar dominant type (31 hands: 24%) had a wide range of skin bifurcation levels, i.e., from 1 to 4, whereas thumbs with the evenly dominant type had more limited variation (all values ≤2.5).

Relationship between skin bifurcation level and nail asymmetry

Nail asymmetry was associated with skin bifurcation levels 0–2, though less frequently at levels 1.5 and 2. At level 0, nails were completely fused at the level of the nailbed and lunula. At level 0.5, nails were partially separated and there was an extension of the proximal nail fold. At level 1 and more proximally, nails were completely separated, with the cleavage extending beyond the level of the germinal matrix. This cleavage was accompanied by cutaneous bridging between the lateral and proximal nail folds, resulting in an overall asymmetrical appearance of the nail. This asymmetry decreased with more proximal skin bifurcation levels. In addition, within subtypes with the same skin bifurcation level, nail asymmetry seemed to decrease as the radial thumb duplicate became more hypoplastic (Online Supplementary Figure 1).

Relationships between skin bifurcation level, joint deviation, and associated osseous asymmetry

Joint deviation and associated deformations of the proximal phalanx were evaluated for the non-floating type, excluding types I and II (Figure 6). Proximal phalangeal head asymmetry was associated with skin bifurcation levels of around 1–2.5 (Figure 6a), whereas proximal phalangeal metaphysis asymmetry had a narrower distribution, with skin bifurcation levels of around 1–1.5 (Figure 6b). The IP joint deviation was strongly associated with skin bifurcation levels around 1–2 (Figure 6c), which had a similar distribution to the proximal phalangeal head asymmetry. The MCP joint deviation had a bimodal distribution, at skin bifurcation levels 1.5 and 4, especially in type IV patients with the ulnar development type (Figure 6d), whereas deviation was infrequent at skin bifurcation levels 2–3. Deviation was frequent in type V (especially at skin bifurcation levels 3 and 4.5).

Figure 7 shows a series of radiographs demonstrating morphological changes of the phalanges depending on the skin bifurcation level. Asymmetry of the proximal phalanx in thumbs with skin bifurcation levels 1–2.5 was characterized by elongation of the epiphysis and tilted morphology of the head. Dispersion of the proximal phalanges in the transverse direction and transverse widening of the metacarpal head were observed, especially in subtypes with more proximal skin bifurcation levels. An association was found in Type IV between the skin bifurcation level and skeletal bifurcation sublevel; more proximal skin bifurcation was correlated with a more proximal location of the phalangeal base of the radial thumb (Online Supplementary Figure 2). Asymmetrical deformations of the proximal phalanx of the ulnar thumb were also observed in the floating type, especially patients with more distal skin bifurcation levels (≤ 2.5) (Online Supplementary Figure 3).

Since nail and skeletal morphologies vary depending on the level of skin bifurcation, it was unsurprising that morphological similarity was found in patients with the same duplication range (Figure 8). Interestingly, morphological changes were very similar for skin bifurcation levels from 1.5 to 3/3.5/4 in Type IV and from skin bifurcation levels from 3 to 5/5.5 in Type VI. The inter-rater reliability was high (Fleiss' Kappa, 0.90) for duplicated thumbs with a non-floating type.

DISCUSSION

In this study, we explored the concept of the duplication range in thumb polydactyly based on the combination of both skin and skeletal bifurcation levels. We were able to correlate proximal phalangeal head asymmetry to predominantly levels 1-2.5 whereas proximal phalangeal metaphysis asymmetry tends to be associated with more distal skin bifurcation levels of 1–1.5. IP joint deviation was found to be strongly associated with levels around 1–2. Finally, MCP joint deviation was associated with two levels: 1.5 and 4.

In thumb polydactyly, preoperative deviation is a risk factor for postoperative deformity, particularly for Wassel type IV thumbs. Hung et al. (1996) classified type IV patients into four subtypes: hypoplastic, ulnar deviated, divergent, and convergent, where the convergent type has a zig–zag morphology in the ulnar thumb, caused by angulations at the IP and MCP joint. Intraoperatively, corrective osteotomy of the first metacarpal is usually recommended, with the amount of correction dependent on the degree of deviation (Hong et al., 2021; Kim et al., 2017; Lee et al., 2013; He and Nan, 2016). Also, tendon centralization is paramount to reduce recurrence, especially of the FPL tendon, which is the main cause of deviation in the convergent type (Hung et al., 1996; Lee et al., 2013). The ulnar-deviated type in the Hung classification (1996) has an ulnar thumb with a less deviated IP joint and an ulnarly deviated MCP joint, and from our study, we were able to reclassify Hung's convergent and ulnar-deviated types by skin bifurcation level. We were also able to further correlate the association between MCP joint deviation with the duplication range; we found a greater splaying of the proximal phalanges and transverse widening of the metacarpal head in subtypes with more proximal skin bifurcation levels (around 2.5 to 4). In addition, we found varying levels of duplication at the MCP joint, with a more proximal location of the phalangeal base of the radial thumb duplicate correlated with more proximal duplication levels. Such knowledge may allow us to further predict the surgical findings preoperatively in order to facilitate planning of the level of osteotomy and other joint levelling procedures.

The premise of our study came from observations that soft tissue abnormalities may be the cause of deformities in thumb duplication. We previously reported a patient with IP joint deviation and proximal phalangeal asymmetry, despite having a very hypoplastic type of thumb duplication (Saito and Morimoto, 2021). We subsequently found a malalignment of the FPL tendon, and in this case, both the deviation and phalangeal deformity were corrected by centralization of the tendon. The presence of deviation and phalangeal deformity, even in severely hypoplastic thumbs, have led us to explore the concept that every phenotype of thumb polydactyl contains underlying soft-tissue abnormalities and carries an inherent risk of deformation. We therefore decided to comprehensively classify our thumb duplication series according to the level of syndactyly so as to provide more prognostic information on the risk of deformity. This has formed the basis of our study and the concept of the duplication range.

There are several soft tissue abnormalities that could account for the deviation of thumb duplicate; other than tendon abnormalities, other variations should be addressed, such as the pulley systems and muscular insertions. In the thumb, the A1 and A2 pulleys are mobile and connect to the volar plate, while the Av and oblique pulleys are immobile and attach to the proximal phalanx (Schubert et al., 2012). Abnormal pulleys have been well documented, especially in type IV patients with convergent morphology and depending on their anatomical sites, these abnormalities vary from conjoined pulleys covering the FPL tendon (Tien et al., 2007) to tough ligamentous membranes bridging the tendon sheaths of the proximal phalangeal heads (He et al., 2017). From both anatomical and biomechanical perspectives, the link between morphological variations in deviation and the skin bifurcation level can be logically deduced (Supplementary Figure 4). For instance, duplication range 1–4 involves all the pulleys, except A1 (level 4) whereas duplication range 3–4 involves all pulleys except

A2 (level 2). While the involved pulleys are duplicated or transversely elongated, the pulleys outside the duplication range may remain independent. This concept is consistent with the finding that IP joint deviation decreased at skin bifurcation levels 2.5 and disappeared at more proximal levels (Figure 6c), where the A2 (level 2) is outside the duplication range.

From the perspective of muscular biomechanics, there was an increased frequency of MCP joint deviation in type IV which can be further distinguishable by skin bifurcation levels. More proximal duplications, e.g., skin bifurcation levels 3.5 and 4, where the A2 pulley (level 2) is outside the duplication range, may be associated with increased integrity of the A2 pulley and therefore a straighter morphology of the phalanges. In addition, splaying of the proximal phalanges and subsequent adductor-dominant muscular imbalance might be a cause of ulnar deviation of the MCP joint. Radial deviation at the IP joint and proximal phalangeal head asymmetry were confined to skin bifurcation levels 1–2 and 1–2.5, respectively. Patients with skin bifurcation levels 1–2.5 correspond to subtypes with a convergent morphology. Asymmetric proximal phalangeal metaphysis was associated with skin bifurcation levels 1–1.5. This finding suggests that patients with convergent morphology can be differentiated by skin bifurcation levels 1–1.5 versus 2–2.5. By contrast, deviations at the MCP joints had a bimodal distribution at around levels 1.5 and 4. Patients with skin bifurcation level 4 correspond to subtypes with an ulnar deviated morphology. Also, patients with skin bifurcation levels 3–3.5 showed an intermediate morphology between the convergent to ulnar deviated types. These findings indicate that the duplication range can potentially be used to grade the morphological characteristics in type IV thumb duplications (Figure 8).

There are limitations to this study. We did not evaluate the agreement between surgical findings and the anatomical abnormalities assumed from the proposed duplication range, as we did not think surgical exploration would necessarily search for all the hypothetical anatomical abnormalities. The relationship between the proposed classification method and postoperative results was not investigated because of the presence of multiple surgery-related variables, which may have complicated the study.

In conclusion, the duplication range concept based on the levels of skin and skeletal bifurcation could potentially increase our understanding of morphology variation in thumb polydactyly. Clinical studies using this classification concept may provide detailed information on risk factors for deformities after reconstructive surgery. This tool can be used not only for decision making regarding surgical options but also for accurately assessing surgical outcomes in radical polydactyly.

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Figures

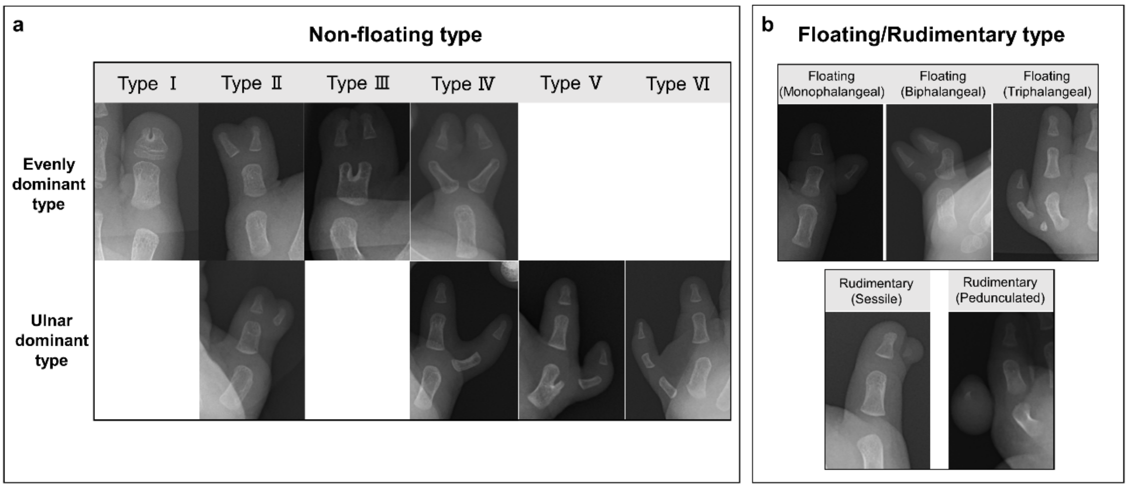


Figure 1. Stratification of thumb duplications by radiological appearance according to the Wassel classification and further sub-classification according to (a) the non-floating type and (b) the floating/rudimentary type.

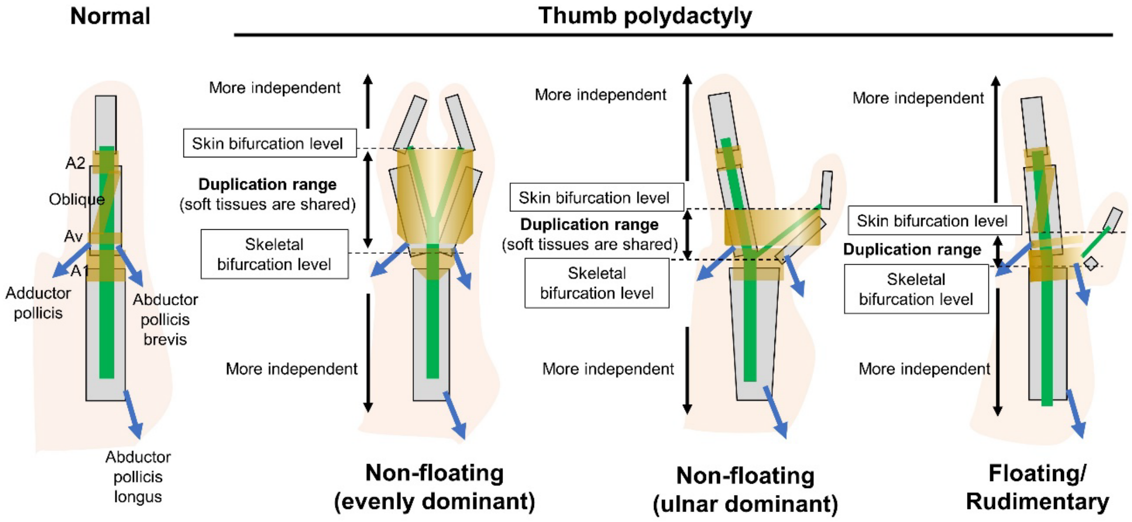


Figure 2. Schematic diagrams to illustrate the duplication range concept. In the syndactylous area between the levels of skin and skeletal bifurcation, abnormally duplicated soft tissues are shared by the two thumbs. These structures are maximally shared in the area, whereas they are more independent proximally and distally. Av, variable annular pulley.

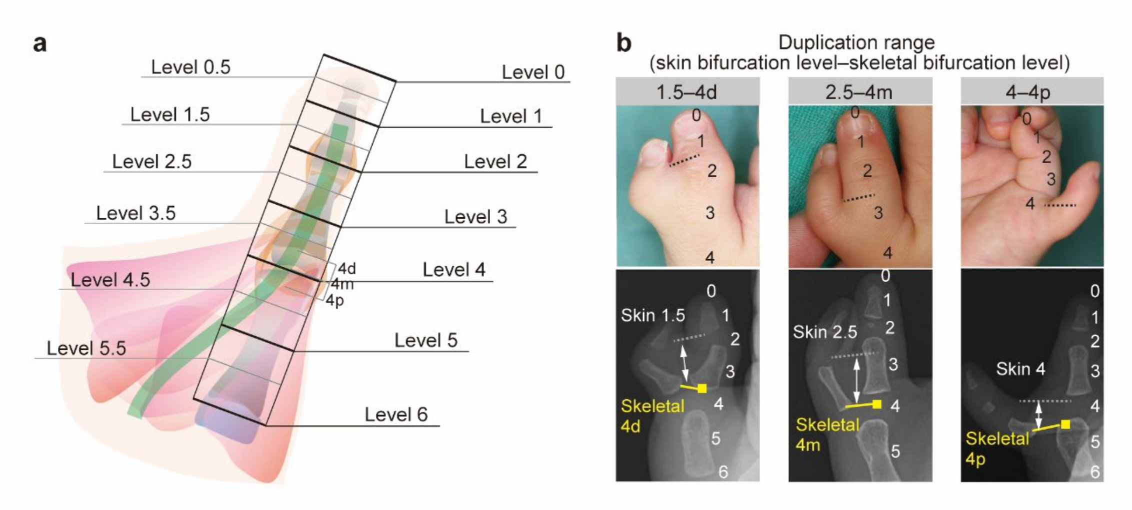


Figure 3. Definitions of skin and skeletal bifurcation levels. Levels are defined along the longitudinal axis of the ulnar thumb duplicate and range from 0 (the tip) to 6 (the carpometacarpal joint) at intervals of 0.5 (**a**). Levels 2 and 4 correspond to the interphalangeal and metacarpophalangeal joints, respectively. Odd-numbered levels are located at the diaphysis of the phalanges and metacarpal. Levels with 0.5 values were defined as the midpoints of neighbouring levels. Three sublevels (4d, 4m, 4p) were defined for the metacarpophalangeal joint (b).

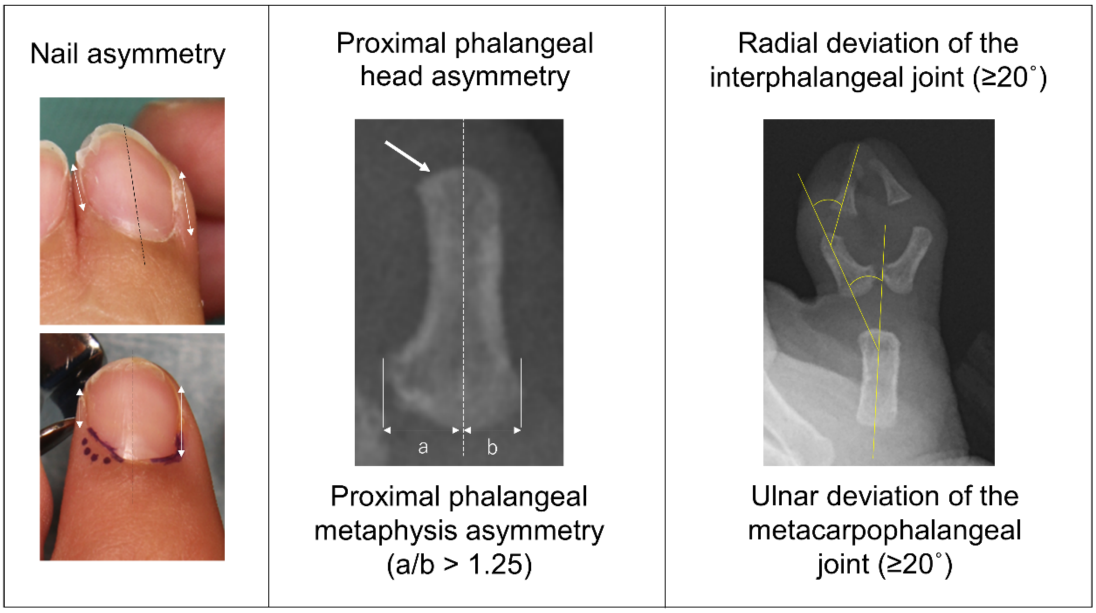


Figure 4.

Morphological parameters used in this study. Nail asymmetry was defined by a deformed lunula, tilted proximal nail fold, or uneven length of the lateral nail folds. Proximal phalangeal metaphysis asymmetry was defined as an asymmetrically elongated base. For this definition, the proximal phalanx was divided into radial and ulnar parts by a longitudinal axis passing through the centre of the diaphysis, and asymmetry was defined as a radial–ulnar ratio of the basal width >1.25. Deviation was diagnosed when the joint deviated at an angle ≥20°.

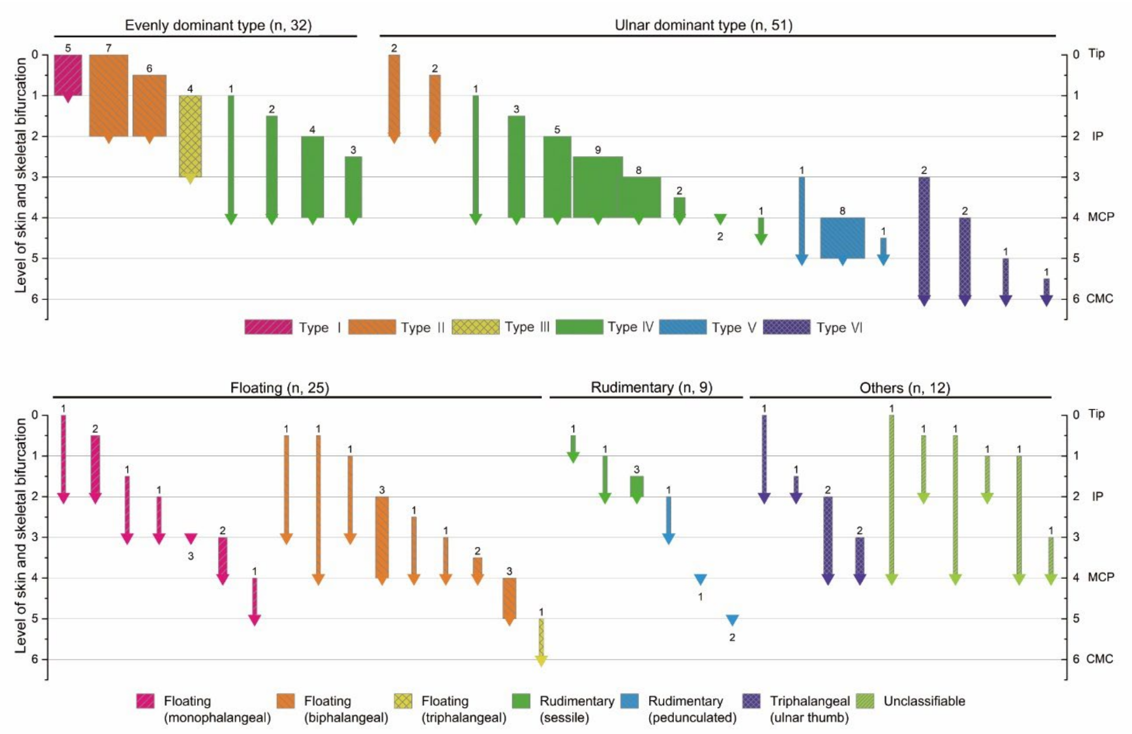


Figure 5. Distribution of non-floating (top) and floating/rudimentary (bottom) thumb polydactyly based on the duplication range concept. The thickness of the bar represents the number of thumbs included in each subtype. There was an increased incidence of types II duplication in the evenly dominant types and type IV duplication in the ulnar dominant types. IP, interphalangeal joint; MCP, metacarpophalangeal joint; CMC, carpometacarpal joint.

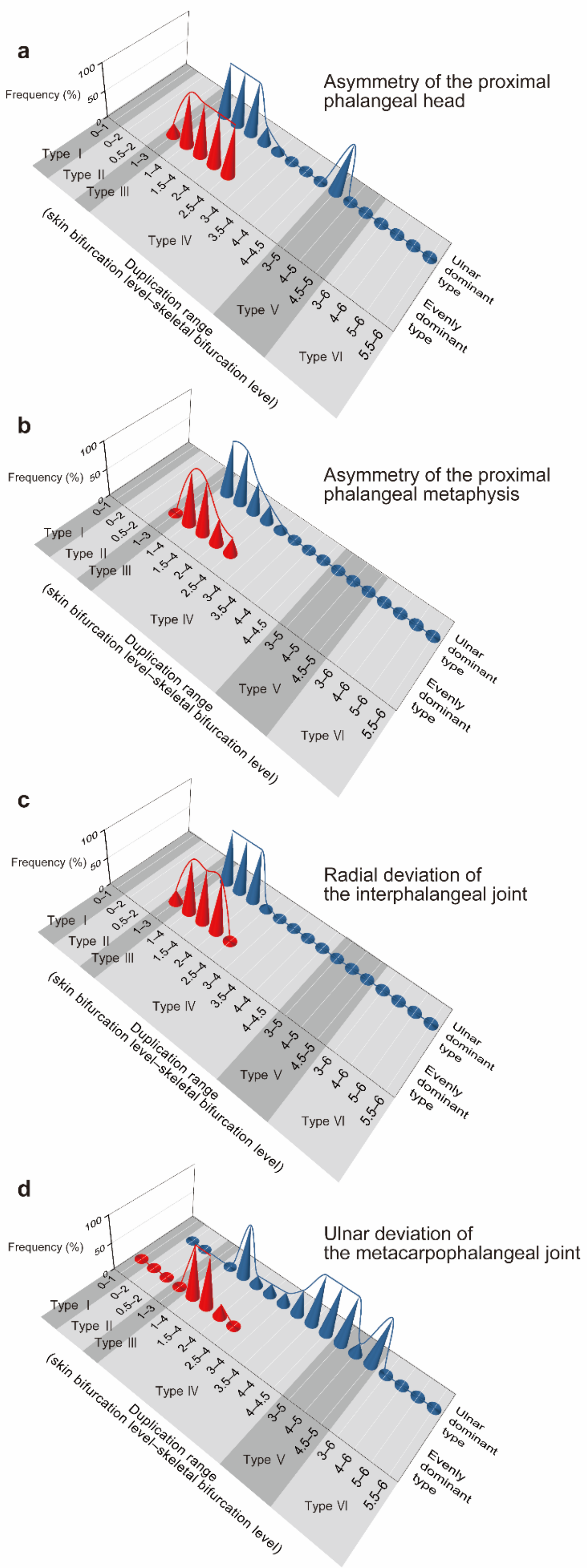

a
Asymmetry of the proximal phalangeal head
b
Asymmetry of the proximal phalangeal metaphysis
c
Radial deviation of the interphalangeal joint
d
Ulnar deviation of the metacarpophalangeal joint
Frequency (%)
Type Ⅰ
Type Ⅱ
Type Ⅲ
Type Ⅳ
Type Ⅴ
Type Ⅵ
Ulnar dominant type
Evenly dominant type
Duplication range
(skin bifurcation level–skeletal bifurcation level)

Figure 6. Relationship between the duplication range and morphological parameters. Cone graphs showed the percentage frequency distribution of proximal phalangeal head symmetry (**a**), proximal phalangeal metaphysis asymmetry (**b**), interphalangeal joint radial deviation (**c**), and metacarpophalangeal joint ulnar deviation (**d**). The cones are shown with spline curves.

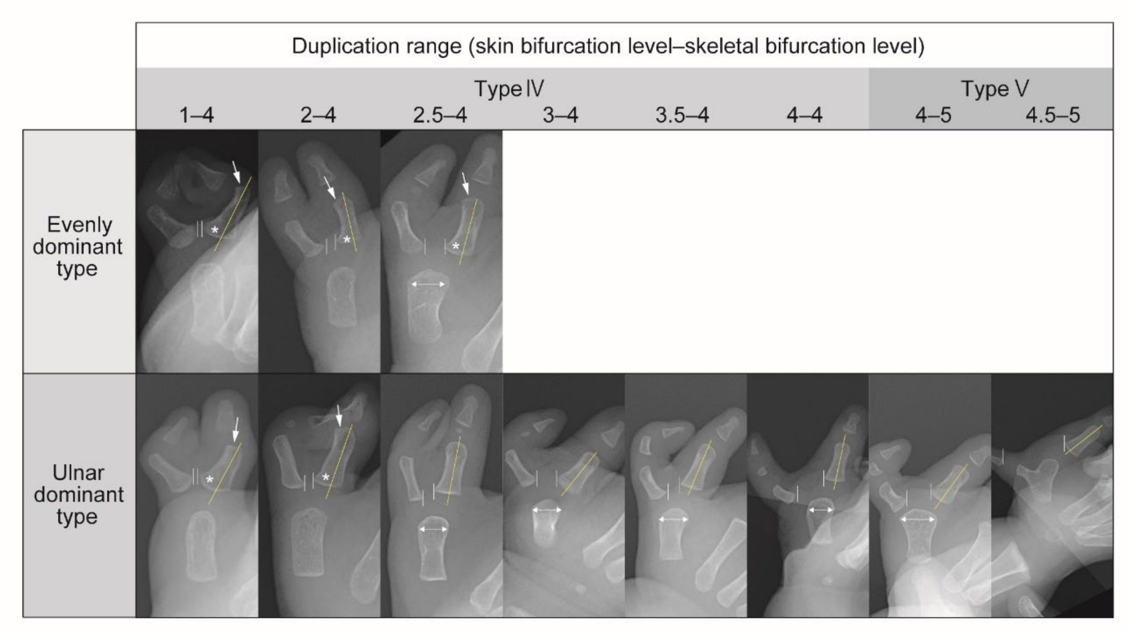


Figure 7.

Variations of skeletal morphology in type IV and V thumb polydactyly according to duplication range. All radiographs are shown with the axis of the first metacarpal aligned in the longitudinal direction. Phalangeal asymmetry is characterized by the linearly tilted morphology of the head (arrows) and uneven protrusion of the proximal metaphysis (stars). Yellow lines indicate the axis of the proximal phalanx. Note that the deformity characterized by divergent proximal phalanges and convergent distal phalanges at skin bifurcation level 1 changes to a morphology with a less deviated metacarpophalangeal joint at skin bifurcation levels 2–2.5, and further progresses into deformity characterized by divergent proximal phalanges with a straightened interphalangeal joint of the ulnar thumb. White lines and bilateral arrows indicate greater dispersion of the proximal phalanges and widening of the metacarpal head.

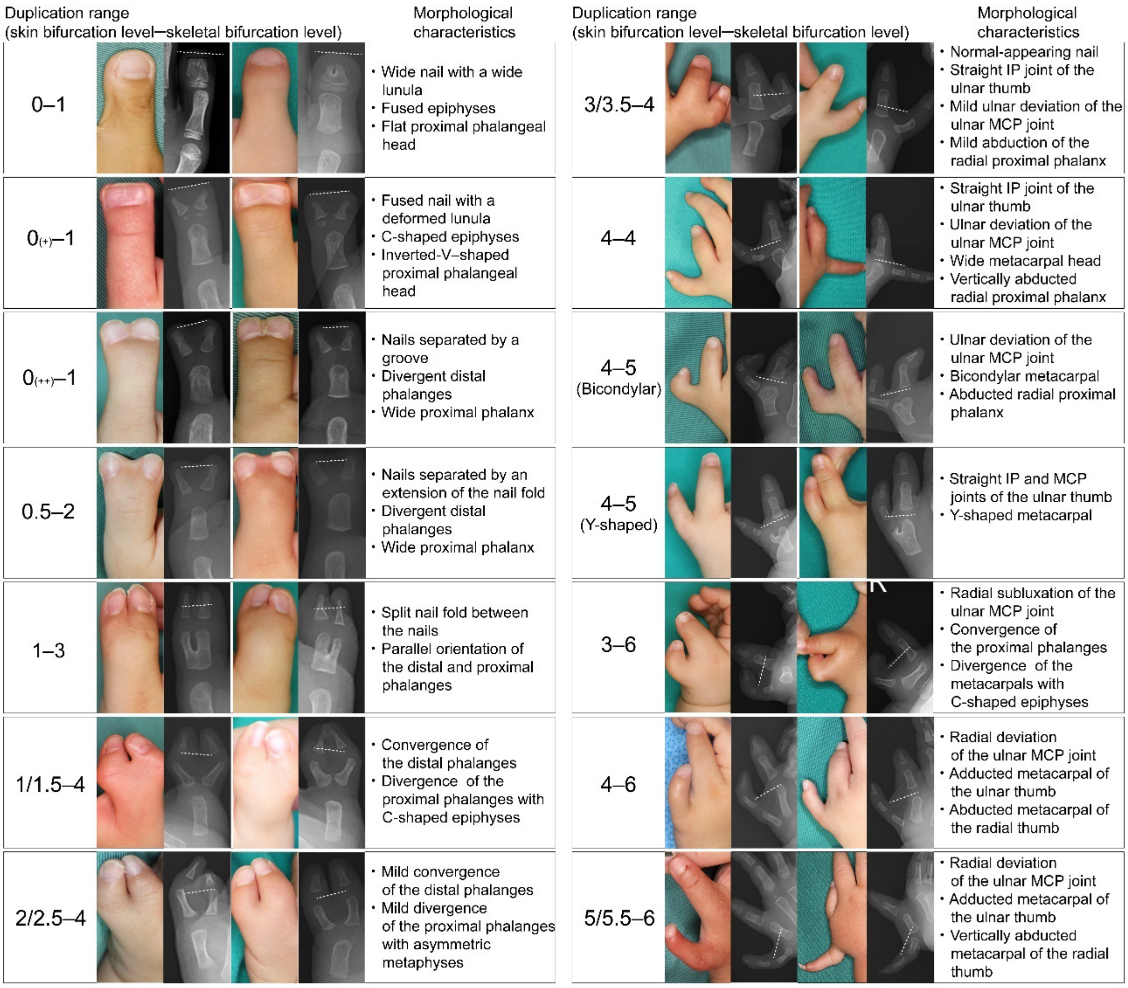
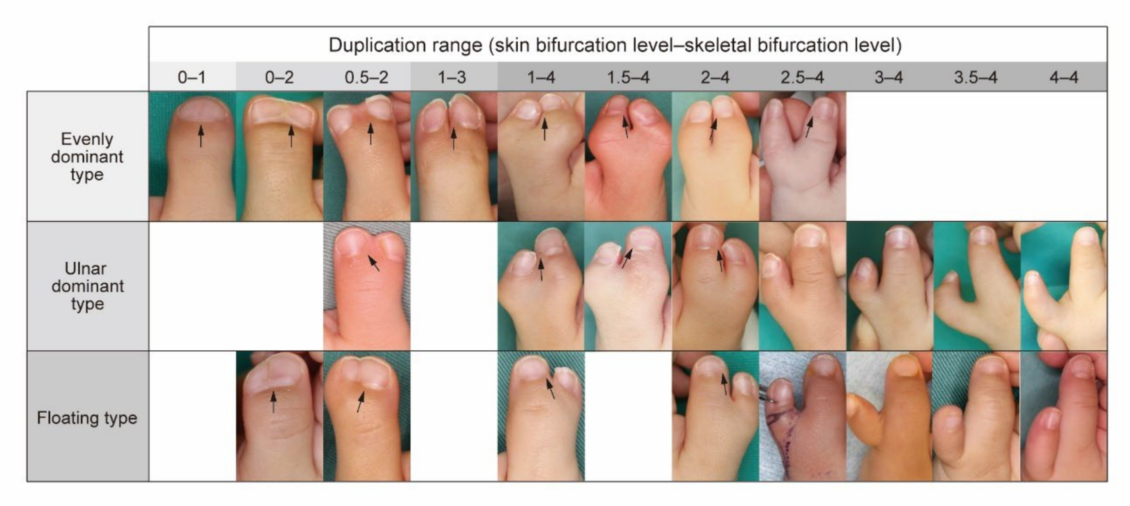
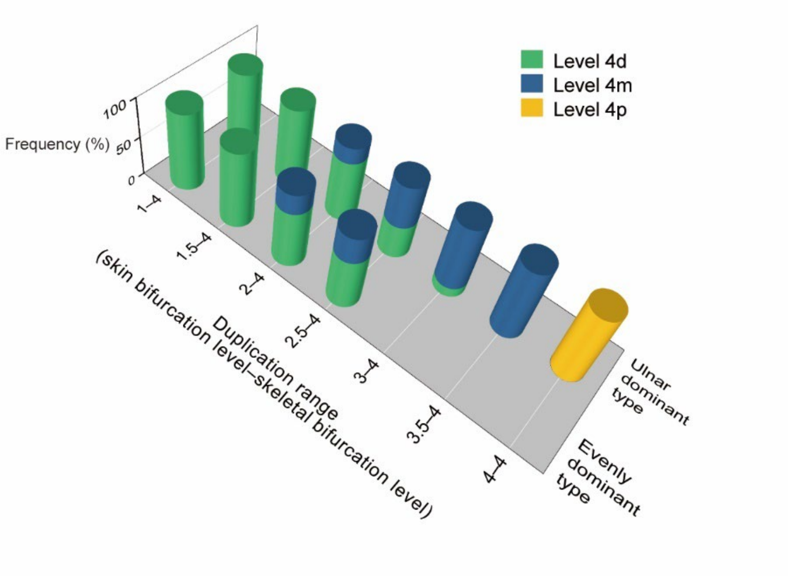


Figure 8.

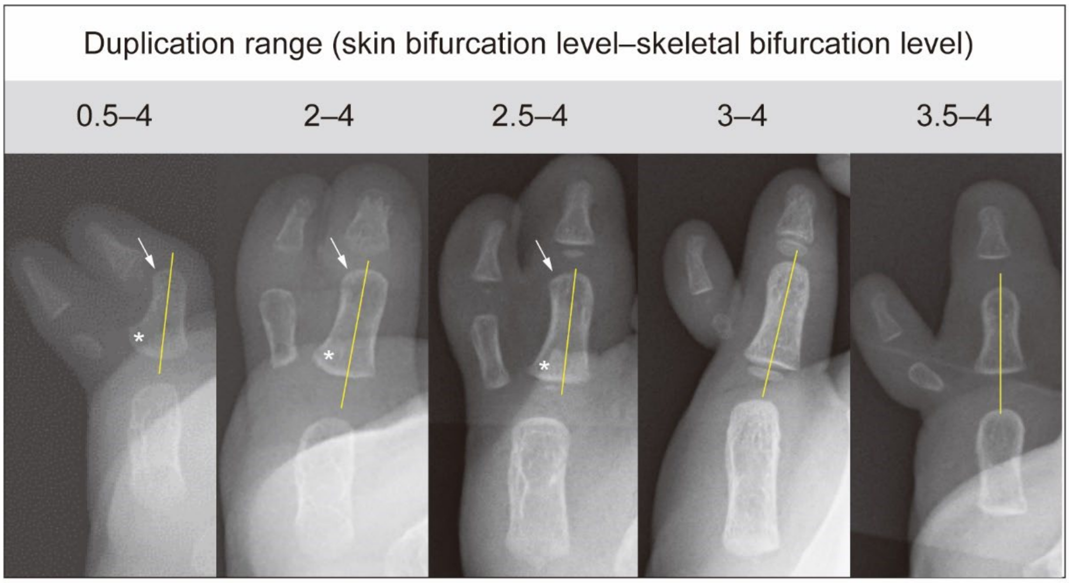
Diagram showing the morphological similarity between thumbs with the same duplication range subtype. Representative morphological characteristics are described for each subtype. Dashed lines represent the level of skin bifurcation. IP, interphalangeal; MCP, metacarpophalangeal.



Online Supplementary Figure 1. Variation of nail asymmetry in thumb polydactyly. Photographs show a representative nail appearance for each subtype. Nail asymmetry is characterized by a deformed lunula or abnormal extension of the proximal nail fold (arrows). Note that nail asymmetry is confined to skin bifurcation level 2 or more distal, especially in the non-floating type.

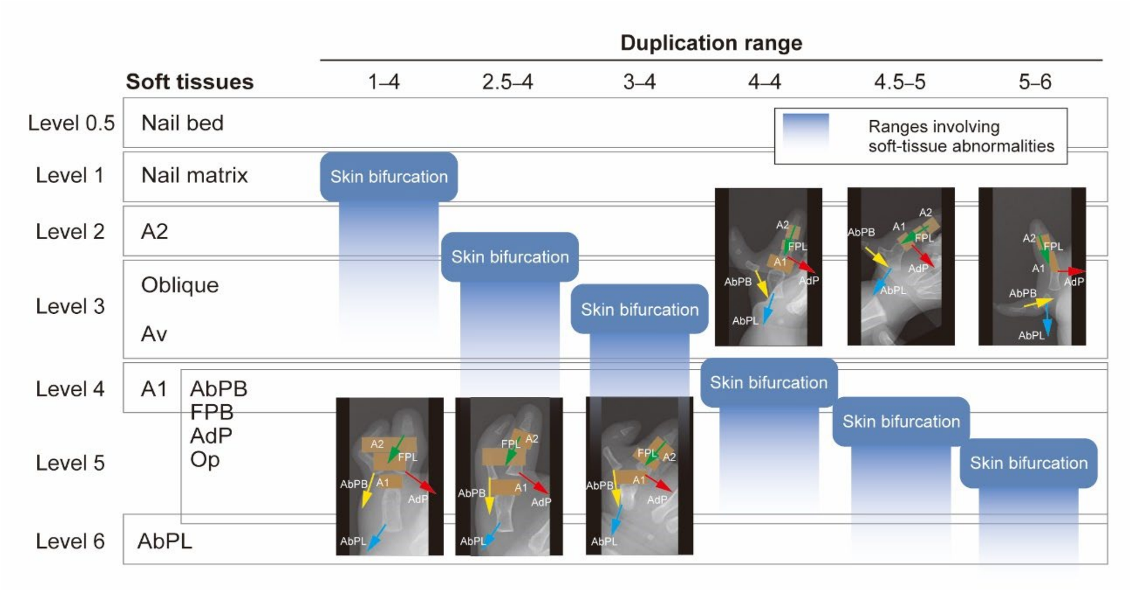


Online Supplementary Figure 2.
Relationship between the level of skin bifurcation and detailed level of skeletal bifurcation for type IV thumb polydactyly. Cylindrical bars represent percent distribution of three sublevels (4d, 4m, and 4p) for level 4.



Online Supplementary Figure 3.

The variation of skeletal morphology in thumb duplications with a non-articulated ulnar thumb, especially those with the biphalangeal type and skeletal bifurcation level 4. Phalangeal asymmetry is characterized by the linearly tilted morphology of the head (arrows) and uneven protrusion of the proximal epiphysis (stars).



Online Supplementary Figure 4. Schematic illustrations showing variations in the involvement of soft tissues depending on duplication range and mechanisms associated with deformation. AdP, adductor pollicis; Av, variable annular pulley; AbPB, abductor pollicis brevis; AbPL, abductor pollicis longus; FPB, flexor pollicis brevis; FPL, flexor pollicis longus; OP, opponens pollicis.