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Ulnar polydactyly of the hand: a classification system and clinical series

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Abstract

Background Ulnar polydactyly, a common congenital hand anomaly, exhibits significant phenotypic variability. Existing classification systems have limitations, particularly in categorizing rare variants. This study introduces a new classification system for ulnar polydactyly that addresses these limitations.

Methods We retrospectively reviewed the medical records of 35 patients with ulnar polydactyly treated at our institution between 2010 and 2022. Data collected included patient demographics, clinical presentation, radiographic findings, family history, associated anomalies, and surgical procedures. Based on detailed morphological and radiographic assessments, we developed a novel classification system comprising five main types (0–4) and associated subtypes.

Results The 35 patients (23 males, 12 females) had a mean age of 3.2 years. Thirty patients had bilateral involvement, with 16 exhibiting symmetry. In total, 65 hands were affected, and one hand was excluded because the patient underwent surgery in another hospital. Type 0 was the most common (38 cases), followed by Type 4 (19 cases), Type 3 (4 cases), and Type 1 (3 cases). Our classification system effectively categorized all cases, including rare variants such as Type 1b (duplicated distal phalanx) and Type 4d (duplication originating from the deformed fourth metacarpal), which are not adequately addressed by previous classifications.

Conclusions Existing classification systems for ulnar polydactyly omit two key variants: Type 1b (duplicated distal phalanx) and Type 4d (duplication from the deformed fourth metacarpal). Our system specifically incorporates these types, providing a more comprehensive framework to guide diagnosis and improve surgical planning for these rare conditions.

Trial registration Retrospectively registered.

Keywords Ulnar polydactyly, Classification, Distal phalanx, Metacarpal bone

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Background

Polydactyly, one of the most common congenital anomalies of the upper limb, can be subdivided into radial, central, and ulnar according to the location of the duplicated digits. Each type of polydactyly usually has one or more specific classifications [1]. Ulnar polydactyly, also known as postaxial polydactyly, refers to disorders with duplication of digits on the ulnar side of the hand. Although the exact mechanism remains unknown, it is thought to be associated with the malfunction of differentiation of the anterior to posterior axis of the hand plate during the development of the upper limb [2]. Ulnar polydactyly may occur as a part of a syndrome or as an isolated event, and many related genes have been reported in the literature [3, 4].

The manifestation of ulnar polydactyly may differ considerably from case to case, and the shape varies from a fully formed finger to a single phalanx or skin nubbin. Several classifications [5–11] (Table 1) methods have been proposed for this purpose. Previous classifications failed to incorporate rare ulnar polydactyly variants, including those with a duplicated distal phalanx and

Table 1 Previously classification in ulnar polydactyly

Temtamy and McKusick Classification [5]	Type A: a fully developed and functioning digit Type B: an incompletely formed and nonfunctioning digit that may be a nubbin or a pedunculated postminimi
Stelling and Turek Classification [6]	Type 1: a digit containing only soft tissue Type 2: a digit containing phalanx Type 3: a fully duplicated digit containing phalangeal and metacarpal
Rayan-Frey Classification [7]	Type I: a cutaneous nubbin without a nail or bone Type II: a pedunculated nonfunctioning digit Type III: a better-developed digit, which articulates with a bifid fifth metacarpal and frequently has a hypoplastic proximal phalanx or not Type IV: a fully developed sixth digit with its own metacarpal Type V: others; including polysyndactyly and other bony abnormalities
Al-Qattan Modification of Rayan-Frey Classification [8]	Type I: a soft tissue nubbin without bone or nail Type II: a pedunculated non-functioning digit with a narrow (< 3 mm) pedicle (type IIA) or a wider (> 3 mm) pedicle (type IIB) Type III: a well-developed digit which articulates with a bifid metacarpal or a partially duplicated metacarpal (type IIIA) or has a fusion between proximal phalanx and the fifth metacarpal (type IIIB) Type IV: a fully developed sixth digit with its own metacarpal Type V: others; including polysyndactyly and triplication of little finger
Pritsch et al. Modified Classification for Type A Ulnar Polydactyly [9]	Type 1 (metacarpal type): a fully developed sixth digit which articulates with carpal Type 2 (metacarpophalangeal type): an extra digit on the lateral side of the fifth digit with an intercalated distal metacarpal remnant Type 3 (phalangeal type): a digit from a hypoplastic sixth metacarpal or a fused fifth metacarpal Type 4 (intercalated type): a digit from metacarpophalangeal joint Type 5 (fully developed type): a digit from a bifid proximal phalanx
Duran et al. Classification [10]	Type I (simple type): a skin nubbin without bone and nail or a nonfunctional digit that contains bone or nail, or both, and a small pedunculated pedicle Type II (hypoplastic type): a digit which presents as a hypoplastic proximal phalanx Type III (proximal phalanx level): Type IIIA: a digit with a bifid proximal phalanx Type IIIB: a digit with a duplicated proximal phalanx Type IV (metacarpal level): Type IVA: a digit which has its fusion between proximal phalanx and metacarpal Type IVB: a digit with a bifid metacarpal head Type IVC: a digit with a metacarpal remnant Type IVD: a digit with a bifid metacarpal or a fully duplicated metacarpal Type V (complicated type): digits which presents as triplication of little finger or polysyndactyly or coexistence of both
Althobaity et al. MAS Classification [11]	For both ulnar and radial polydactyly: Soft tissue attachment Joint attachment: I: Attached to carpometacarpal joint II: Attached to metacarpophalangeal joint III: Attached to proximal interphalangeal joint IV: Attached to distal interphalangeal joint Bone attachment: I: Attached to metacarpal II: Attached to proximal phalanx III: Attached to middle phalanx IV: Attached to distal phalanx

those with duplication originating from the deformed fourth metacarpal. Our classification addresses this gap by categorizing ulnar polydactyly based on the most proximal level of skeletal involvement, progressing from rudimentary digits (Type 0) to those involving the distal phalanx (Type 1), middle phalanx (Type 2), proximal phalanx (Type 3), and metacarpal (Type 4). This approach, mirroring the simplicity of the Wassel classification for radial polydactyly, provides a framework for understanding the extent of the duplication and guiding surgical decision-making.

Materials and methods

The study protocol received ethical approval from the Institutional Review Board of Beijing Jishuitan Hospital (Reference No. 201808-09). Written informed consent was obtained from all participants before inclusion in the study. We retrospectively studied 35 patients diagnosed with ulnar polydactyly who were admitted to inpatient and outpatient clinics between January 2010 and January 2022. Inclusion criteria: ulnar polydactyly diagnosed clinically and radiographically. Exclusion criteria: (1) Medical records with insufficient data for comprehensive analysis, including missing clinical notes or radiographs; (2) Patients who underwent surgical correction of the ulnar polydactyly before enrollment in this study, as this compromises the ability to accurately characterize the initial presentation and anatomy.

Data were extracted independently by two blinded hand surgeons using a standardized form and reviewed for logical consistency by a third hand surgeon. Data included demographics, family/perinatal history, concurrent anomalies, physical exam findings, radiographs, and surgical records.

A classification system was developed iteratively, reviewing existing systems of radial and ulnar polydactyly and prioritizing the level of duplication, and the feature of the duplicated finger. The system includes five types (0–4) and subtypes.

To assess the reliability of the classification system, a random sample of 30 de-identified cases of ulnar polydactyly was presented to three independent hand surgeons from our institution who were not involved in the development of the classification. Each surgeon classified the 30 cases twice, with a two-week interval between assessments. Intra-observer reliability was assessed using Cohen's kappa coefficient for each rater, and inter-observer reliability across all raters was calculated using Fleiss' kappa coefficient. Kappa values were interpreted as follows: >0.80, excellent agreement; 0.60–0.80, substantial agreement; 0.40–0.60, moderate agreement; 0.20–0.40, fair agreement; and <0.20, slight agreement. Statistical analyses were performed using IBM SPSS Statistics Version 27.

Results

In this study, 35 patients with ulnar polydactyly were included: 23 males (65.7%) and 12 females (34.3%). The mean age of the patients was 3.2 years, ranging from 4 months to 18 years. Thirty patients (85.7%) had bilateral polydactyly, and 16 of these bilateral cases (53.3%) were symmetric. This resulted in a total of 65 affected hands, with 31 (47.7%) on the left side and 34 (52.3%) on the right side.

Analysis of our clinical series, we propose a new classification system (Fig. 1) for ulnar polydactyly based on the most proximal skeletal element involved– ranging from rudimentary digits without skeletal connections (Type 0) to duplications affecting the metacarpal (Type 4). This classification allowed us to identify patterns in the anatomical presentations of ulnar polydactyly. It includes five main types and their subtypes, detailed as follows.

The classification system used in the present study

Type 0 The duplicated digit is hypoplastic and lacks a skeletal connection to the hand.

- Type 0a: A nubbin with or without a nail.
- Type 0b: A floating digit connected to the hand by a skin bridge.
- Type 0c: A hypoplastic, nonfunctioning little finger connected to the hand by a non-bone structure on X-ray. The duplicated digit has a similar appearance and structure to a normal little finger but is underdeveloped and lacks independent movement.

Type 1 (distal phalanx type): Duplication or deformity of the distal phalanx.

- Type 1a: Bifid or irregular distal phalanx of the little finger.
- Type 1b: Duplicated distal phalanx of the little finger.

Type 2 (middle phalanx type): Duplication or deformity of the middle phalanx.

- Type 2a: Bifid or irregular middle phalanx of the little finger.
- Type 2b: Duplicated middle phalanx of the little finger.

Type 3 (proximal phalanx type): Duplication or deformity of the proximal phalanx.

- Type 3a: Bifid or irregular proximal phalanx of the little finger.
- Type 3b: Duplicated proximal phalanx of the little finger.

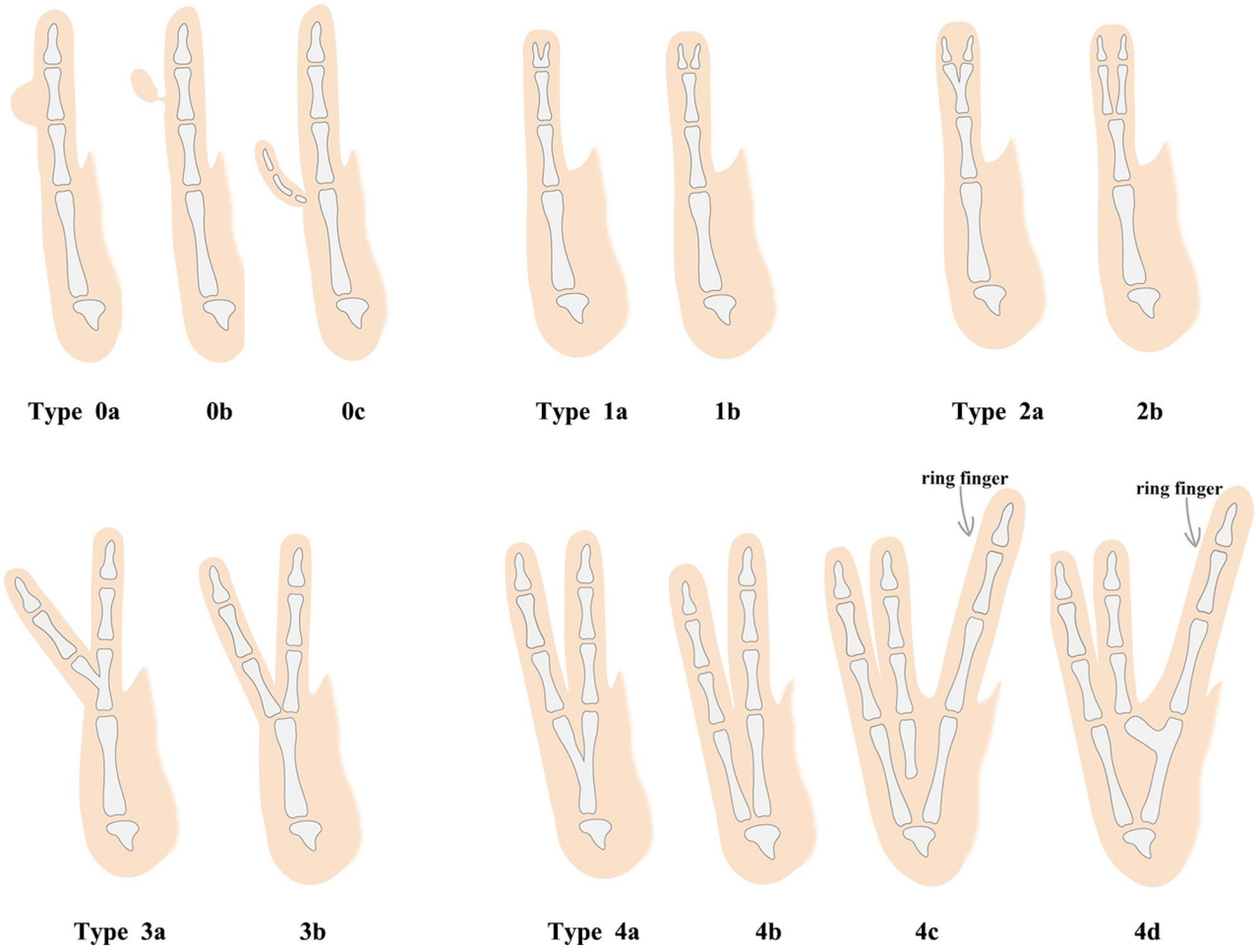

Fig. 1 Schematic diagram of the proposed ulnar polydactyly classification system, illustrating the five main types (0–4) and their subtypes

Table 2 Results of intra-observer agreement

	Kappa Value	Interpretation of agreement
Observe 1	0.814	excellent
Observe 2	0.850	excellent
Observe 3	0.814	excellent

Table 3 Results of inter-observer agreement

	Kappa value	Interpretation of agreement
First assessment	0.703	substantial
Second assessment	0.689	substantial
Overall	0.727	substantial

Type 4 (metacarpal type): Duplication or deformity of the metacarpal bone.

- Type 4a: Bifid or irregular fifth metacarpal.
- Type 4b: A fully developed supernumerary metacarpal that articulates separately with the carpals.
- Type 4c: A duplicated, hypoplastic metacarpal.
- Type 4d: Duplication of the little finger originating from the deformed fourth metacarpal bone.

Intra-observer and inter-observer reliability results are summarized in Tables 2 and 3, respectively. The intra-observer reliability of the classification system was excellent, with all observers demonstrating a high degree of consistency in their classifications. The inter-observer reliability of the classification system showed substantial agreement across all assessments. This indicates a good level of consistency between different observers applying the classification system.

While this classification does not formally include complex cases, such as those with triplicated or more duplicated little fingers, it can still be applied to them. Our classification regards the complex deformity as a combination of several basic malformations. Each basic malformation can be categorized into a specific type in this classification. Therefore, one complex ulnar polydactyly may be classified as having multiple types.

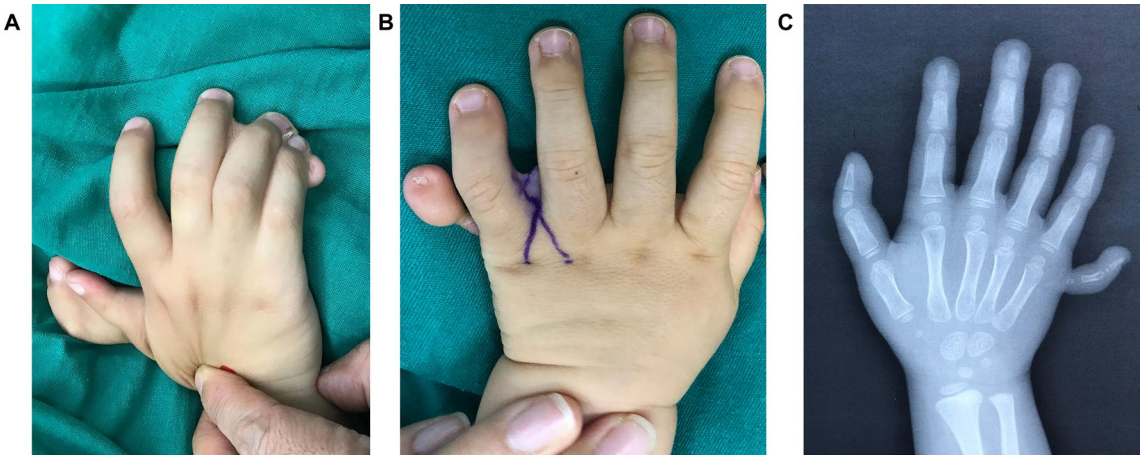

Fig. 2 Clinical and radiographic examples of Type 0 ulnar polydactyly. (**A**) Preoperative view of a Type 0a case (nubbin), also presenting with syndactyly and radial polydactyly. (**B**) Preoperative view of a Type 0b case (floating digit connected by a skin bridge). (**C**) X-ray of a Type 0c case, showing a hypoplastic little finger without a skeletal connection to the hand

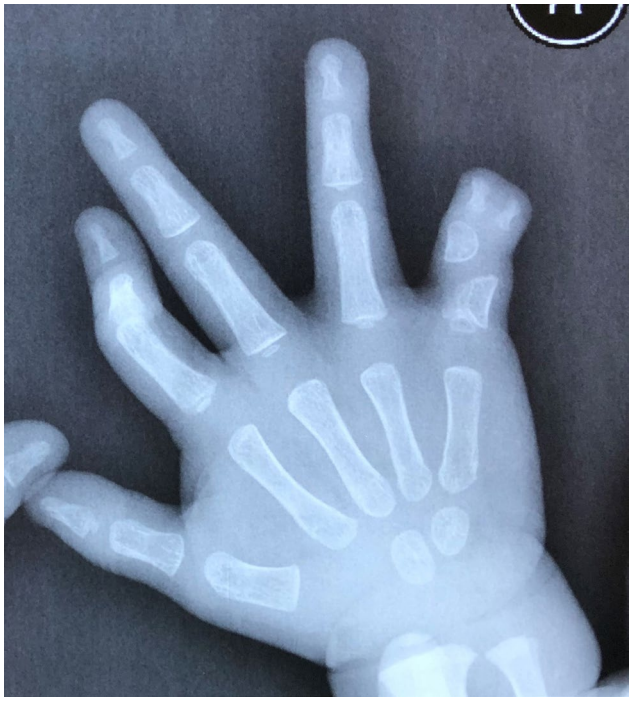

Fig. 3 X-ray of a Type 1b ulnar polydactyly case, showing a duplicated distal phalanx of the little finger. Note the presence of a triangular-shaped proximal phalanx

One patient with bilateral ulnar polydactyly underwent surgery on his left hand at another hospital. Preoperative data for his left hand were incomplete and thus excluded. According to this classification, the distribution of 64 cases was as follows: Type 0 was the most frequent, comprising 38 cases, further categorized into subtypes 0a (6 cases), 0b (16 cases), and 0c (16 cases) (Fig. 2). Type 1 included 3 cases, all of which were subtype 1b (Fig. 3). Type 3 was observed in 4 cases, with 1 case of subtype 3a and 3 cases of subtype 3b (Fig. 4). Finally, Type 4 accounted for 19 cases, subdivided into subtypes 4a (9 cases), 4b (7 cases), 4c (1 case), and 4d (2 cases) (Fig. 5). Rare cases of type 1a and type 4d, which have not been previously reported, were observed in this clinical series.

Furthermore, nine patients had a positive family history of polydactyly of the hand or foot, and five had a family history of syndactyly. Thirty patients in this clinical series had other coexisting congenital diseases. Polydactyly of the foot was the most common, identified in 22 cases, while syndactyly of the hand or foot was observed in 15 cases. Radial polydactyly (4 cases), triphalangism (2 cases), and single instances of thumb hypoplasia, club feet, and radial longitudinal deficiency were also noted. Congenital heart disease was present in three patients, manifesting as atrial septal defect, ventricular septal defect, or arteriovenous ductus arteriosus. One case each of bronchopulmonary dysplasia, developmental delay, and structural brain anomaly were also recorded.

One patient, who had bilateral ulnar polydactyly, polydactyly of both feet, syndactyly of the hand and foot, and structural brain anomalies, underwent a genetic test, which showed that the patient had a heterozygous mutation in the GLI3 gene on chromosome 7 and was diagnosed with Greig cephalopolysyndactyly syndrome, which is an autosomal dominant disorder associated with preaxial and postaxial polydactyly and variable syndactyly [12].

Discussion

Despite the existence of numerous classification schemes for ulnar polydactyly (Table 1), further refinement is needed. The Stelling classification [6] and the Temtamy and McKusick classification [5] were early proposed, and later some more detailed modifications were came up with in the Rayan-Frey classification, Al-Qattan classification, and Pritsch et al. modified classification. The

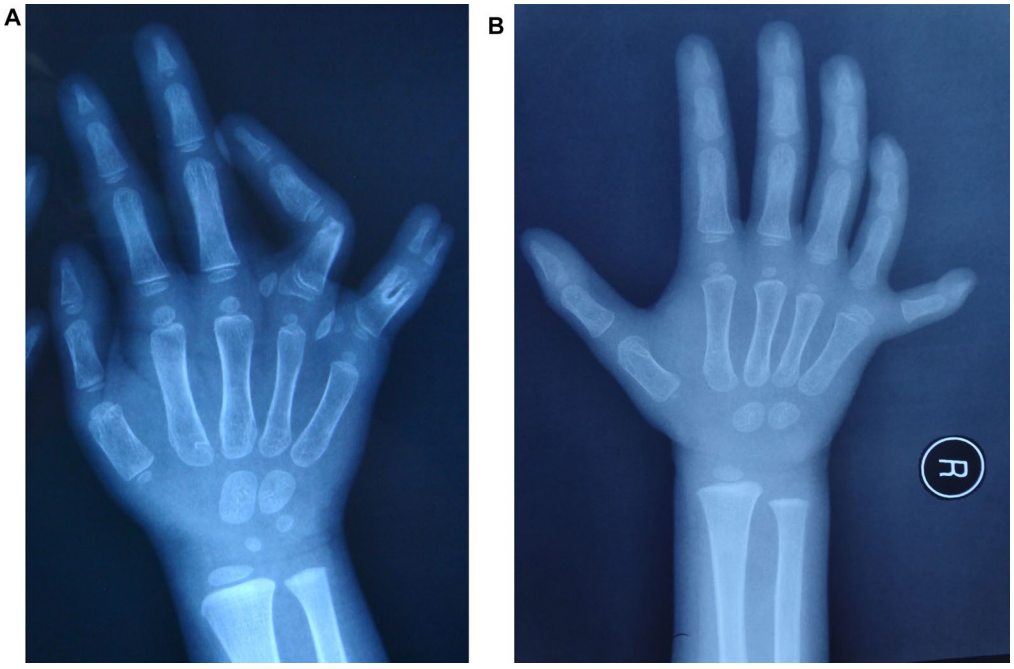

Fig. 4 Radiographic examples of Type 3 ulnar polydactyly. (**A**) X-ray of a Type 3a case, showing a bifid proximal phalanx of the little finger. Note the hypoplastic middle phalanges of the little fingers and the abnormal ring finger with small bones around it. (**B**) X-ray of a Type 3b case, showing a duplicated proximal phalanx

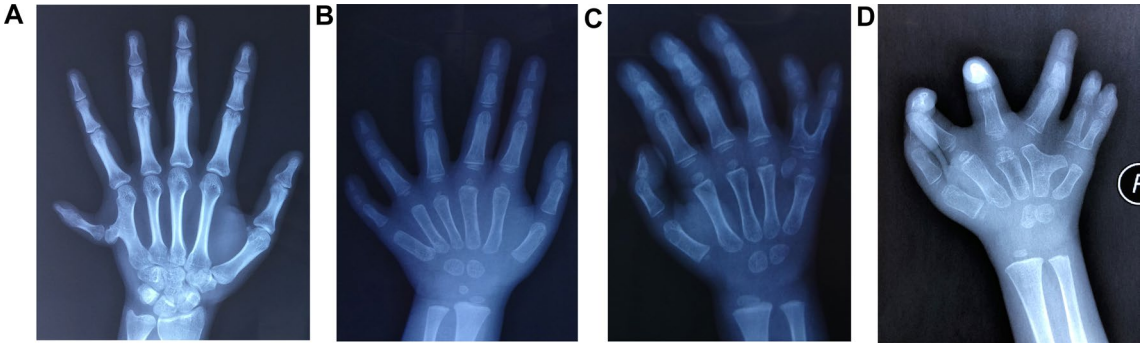

Fig. 5 Radiographic examples of Type 4 ulnar polydactyly. (**A**) X-ray of a Type 4a case, showing a bifid fifth metacarpal. (**B**) X-ray of a Type 4b case, showing a fully duplicated fifth metacarpal. (**C**) X-ray of a Type 4c case, showing a hypoplastic, duplicated fifth metacarpal. Note the fusion between the proximal phalanges of the fifth and supernumerary digits. (**D**) X-ray of a Type 4d case, showing a duplication of the little finger originating from the bifid fourth metacarpal

Rayan-Frey classification [7] and the Al-Qattan classification [8] both defined a mixed type V for all complicated cases, which may group together disparate conditions. Duran et al [10]. developed a complex type V specifically for triplication of the little finger, a rare case of ulnar polydactyly. Pritsch et al [9]. modified type A of the Temtamy and McKusick classification, but the most distal level of the duplication was the proximal phalanx. The MAS classification [11] was an interesting one, which could be applied to both the radial and ulnar polydactyly. Nevertheless, a rare case in our clinical series (Fig. 5D), involving a little finger-like duplication arising from the deformed fourth metacarpal, could not be categorized using the Duran classification, Pritsch et al. modified classification, and the MAS Classification.

In contrast to radial polydactyly, ulnar polydactyly with duplications originating from the phalanges is relatively uncommon [9]. The most distal duplication previously reported involved the middle phalanx [13]. Our clinical series, however, included cases with duplications of both the proximal and distal phalanges. Consequently, we propose types 1, 2, and 3 to encompass these variations, with each type further divided into two subtypes based on the specific characteristics of the phalangeal malformation. Although we did not observe cases of types 1a, 2a, and 2b in our series, AlNojaidi et al [13]. reported one rare case that fits the description of type 2b. We postulate that

types 1a and 2a may also exist and could be identified in future studies; thus, we have included them in our classification. Consistent with previous studies, Type 0 was the most prevalent type in our clinical series. Types 3a and 4c, however, were each represented by a single case. The rarity of these subtypes is notable, although they have been documented in other research. For instance, Pritsch et al [9]. and Duran et al [10]. both reported cases that fit the characteristics of type 3a and type 4c.

Although this classification primarily focuses on simpler forms of ulnar polydactyly, it can be effectively applied to complex cases, including those with triplicated or even more duplicated little fingers. To achieve this, we propose analyzing the complex deformity as a combination of several basic malformations, each of which can be categorized into a specific type within the classification. For example, the triplicated little finger reported by Duran et al [10]. could be interpreted as having two separate instances of Type 3b, each representing a duplicated proximal phalanx. This approach allows for a systematic and concise classification of even the most complex cases.

Surgical treatment is generally the preferred approach for ulnar polydactyly, and our classification system provides a framework for guiding surgical decisions by correlating the anatomical presentation with specific surgical considerations. Types 0a and 0b, which are type B in the Temtamy and McKusick classification, are usually easy to handle through suture ligation or surgical excision [14]. However, complications associated with suture ligation, such as neuroma formation, cyst formation, aesthetically unacceptable residual stumps, and infection, are surprisingly more common than in surgical excision [1]. Therefore, we recommend careful consideration of these potential drawbacks when managing these types. Type 0c presents a unique challenge, as radiographs reveal no bony connection between the radial and ulnar little fingers. However, based on our clinical experience, a cartilaginous connection is consistently observed intraoperatively and must be fully excised to prevent recurrence. For Types 1, 2, and 3, the classification alerts the surgeon to the possibility of synpolydactyly, necessitating careful attention to nail bed management and potential reconstruction of the PIP and/or DIP joints. Furthermore, depending on the specific anatomical variations, reconstruction of the flexor digitorum superficialis and/or flexor digitorum profundus tendons may be required to restore optimal function. Type 4 duplications offer the possibility of preserving the whole finger with better morphology and function, or performing a more complex reconstruction to merge part of the ulnar and radial little fingers into a complete one. In these cases, the classification guides the surgeon to assess the stability of the MCP joint and consider reconstruction of structures such as the abductor digiti minimi, ulnar collateral ligament, and capsule, as well as the deep transverse metacarpal ligament to restrict metacarpal splay. Finally, the presence of bifid or irregular phalanges or metacarpals, as seen in types 1a, 2a, 3a, 4a, and 4d, often necessitates metacarpal or phalanx osteotomy to achieve proper alignment and separation of the extra part.

Additionally, ulnar polydactyly can present as either a single disease or part of a syndrome. Ulnar polydactyly tends to be associated with foot deformities [3], which was also observed in our clinical series. Furthermore, several mutations, such as SHH mutations, MIPOLI, PITXI, GLI3 on chr7p13, and PAPA2 and PAPA3 on chr13q21-q32 and chr19p13.2-p13.1, respectively [3, 4], are thought to be relevant to this malformation. There are approximately 17 common syndromes related to ulnar polydactyly [6]. They can be mainly divided into three groups: first, ciliopathy syndromes due to gene mutations that are associated with the structure of the cilia or intraflagellar protein transport; second, syndromes due to mutations of the GLI3 gene that cause truncations of the Gli3 protein; third, syndromes due to chromosomal duplication, such as Patau syndrome (Trisomy 13) [15]. In this study, one patient was diagnosed with Greig cephalopolysyndactyly syndrome, linked to a heterozygous GLI3 mutation. It is a limitation of this study, as the conditions for family members positive for these mutations were not recorded in detail, and most patients did not undergo a gene sequencing; therefore, it was difficult to explore the genetic relationships among different types.

Compared to these prior systems, our classification offers three key advantages. First, it incorporates previously unreported variations, including rare presentations of Type 1 and Type 4D ulnar polydactyly. Second, the classification is organized based on the most proximal level of skeletal involvement. This organization, analogous to the widely adopted Wassel classification for radial polydactyly, provides a more intuitive and clinically relevant framework for surgeons. Third, our system can be applied to complex cases of ulnar polydactyly, such as triplicated little fingers, by analyzing them as a combination of basic malformations, each classifiable within our framework.

Our study has several limitations that warrant acknowledgement. First, as a single-center retrospective study, the cases are limited, and two proposed subtypes were observed in series. We retained these subtypes to maintain a comprehensive framework, acknowledging the need for future validation with a larger dataset. Second, due to the retrospective nature of this study, a direct assessment of predictive validity was not feasible. However, we aim to address this limitation in future prospective studies by correlating classification type with surgical decision-making, post-operative outcomes, and

long-term functional results. Third, this study does not include a detailed analysis of post-operative outcomes. Future research will focus on evaluating the clinical effectiveness of the classification in guiding surgical treatment and improving patient outcomes.

Conclusions

Current classification systems for ulnar polydactyly do not adequately address two clinically important variants: those with a duplicated distal phalanx (Type 1b) and those with duplication originating from the deformed fourth metacarpal (Type 4d). Our classification system specifically includes these types, offering a more comprehensive framework to guide diagnosis and improve surgical planning for these rare conditions.

Abbreviations

MCP	Metacarpophalangeal
PIP	Proximal interphalangeal
DIP	Distal interphalangeal

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Author contributions

Wen Tian contributed to the design of the work and manuscript revision. Wenyao Zhong, Liying Sun, and Zongxuan Zhao collected and analyzed the patient data. Dan Chen analyzed and interpreted the patient data, prepared all figures, and was a major contributor in writing the manuscript. All authors read and approved the final manuscript.

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Data availability

No datasets were generated or analysed during the current study.

Declarations

Ethics approval and consent to participate

This study was reviewed and approved by the Institutional Review Board of Beijing Jishuitan Hospital (Reference No. 201808-09). Written informed consent was obtained from each patient prior to their participation in the study.

Consent for publication

Not applicable.

Competing interests

The authors declare no competing interests.

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