



Thumb duplication classifications: Is there still a need for improvement?

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About 16% of upper extremity congenital abnormalities consist of thumb defects.^[1] Thumb duplication, also named as preaxial polydactyly or bifid thumb, occurs in 1 per 10,000 births in Caucasians and Asians.^[2] It occurs sporadically; however, rare associated visceral anomalies have been described.^[2] It is usually unilateral. Associated anomalies and positive familial history are more common in bilateral cases.^[3] The range of thumb polydactyly can vary from a bifid distal phalanx to duplication of the radial carpal bones and bifurcated radius or triplicated thumb.^[4,5]

Surgeons have sought an ideal and easily applicable classification system that helps to document the types of deformity and guide

ABSTRACT

Objectives: This study aims to compare the usefulness of two systems in classifying thumb duplication cases and give some examples of the cases we believe that are unclassifiable.

Patients and methods: Between January 2011 and January 2018, a total of 50 patients (29 males, 21 females; median age: 46.4±68.3 months; range, 1 to 318 months) with thumb duplications as assessed according to the Wassel and Rotterdam classification systems were included.

Results: Duplication was present in the right hand in 28, in the left hand in 21, and in both hands in one patient. According to the Wassel classification system, 45 patients could be allocated in any of the types; however, five patients could not be classified. According to the Rotterdam classification, 47 cases fell into one of the classifications; however, three cases could not be classified.

Conclusion: Despite efforts to find the best classification system for thumb duplications, the proposed systems may not fully cover the presented radial polydactyly cases, and additions to the system are required.

Keywords: Classification, hand deformities, thumb, polydactyly.

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treatment. The Wassel classification system is the most often used system owing to its simplicity in defining the deformity.^[6] Wassel type IV, which is the complete duplication of the proximal and distal phalanges, is the most common type of deformity, followed by type II, a completely duplicated distal phalanx, and type VII, a triphalangeal thumb or elements of a triphalangeal thumb accompanied by a normal thumb. Some modifications have been made, as the Wassel classification system has certain drawbacks, such as insufficiency to classify all spectra of anatomical complexity, and it barely helps in surgical planning.^[7]

The Rotterdam classification system, which combines the Wassel system and Buck-Gramcko and Behrens intercarpal modification, is proposed to indicate different complex deformities, such as triphalangism, triplication, symphalangism, deviation, and hypoplasty.^[8] To help in surgical planning, Chung et al.^[9] divided cases into four categories: joint type, epiphyseal type, osteochondroma-like type, and hypoplastic type.

All classifications must be useful for communication within the literature, clinical care, and formation of registries, as well as help in surgical decision making. Despite the efforts to categorize congenital hand deformities based on their morphologic or radiological characteristics, basic reconstruction principles are still used to achieve optimal results.^[10] Some reports have presented cases that cannot be easily distinguished by Wassel or Rotterdam classification.^[11-13] In the present study, we, therefore, aimed to compare the applicability of these two most commonly used classification systems in our thumb duplication cases and to define whether any cases were unclassifiable by either scheme.

PATIENTS AND METHODS

This two-center, retrospective study was conducted at Hand Surgery Clinics of Metin Sabancı Baltalimanı Bone Diseases Training and Research Hospital and Adana City Training and Research Hospital between January 2011 and January 2018. A total of 50 patients with thumb duplications (29 males, 21 females; median age: 46.4±68.3 months; range, 1 to 318 months) were included. A written informed consent was obtained from the parents and/or legal guardians of the patients and the patient (Case 2). The study protocol was approved by the Metin Sabancı Baltalimanı Bone Diseases Training and Research Hospital Institutional Review Board (IRB No. 2018/36-245). The study was conducted in accordance with the principles of the Declaration of Helsinki.

Posteroanterior and lateral radiographs of the duplicated thumb were obtained at the outpatient clinic at the last visits of the patients, preoperative ones if the patient was operated, from the Picture Archiving and Communication Systems (INFINITT PACS®, INFINITT Healthcare, Seoul, South Korea)

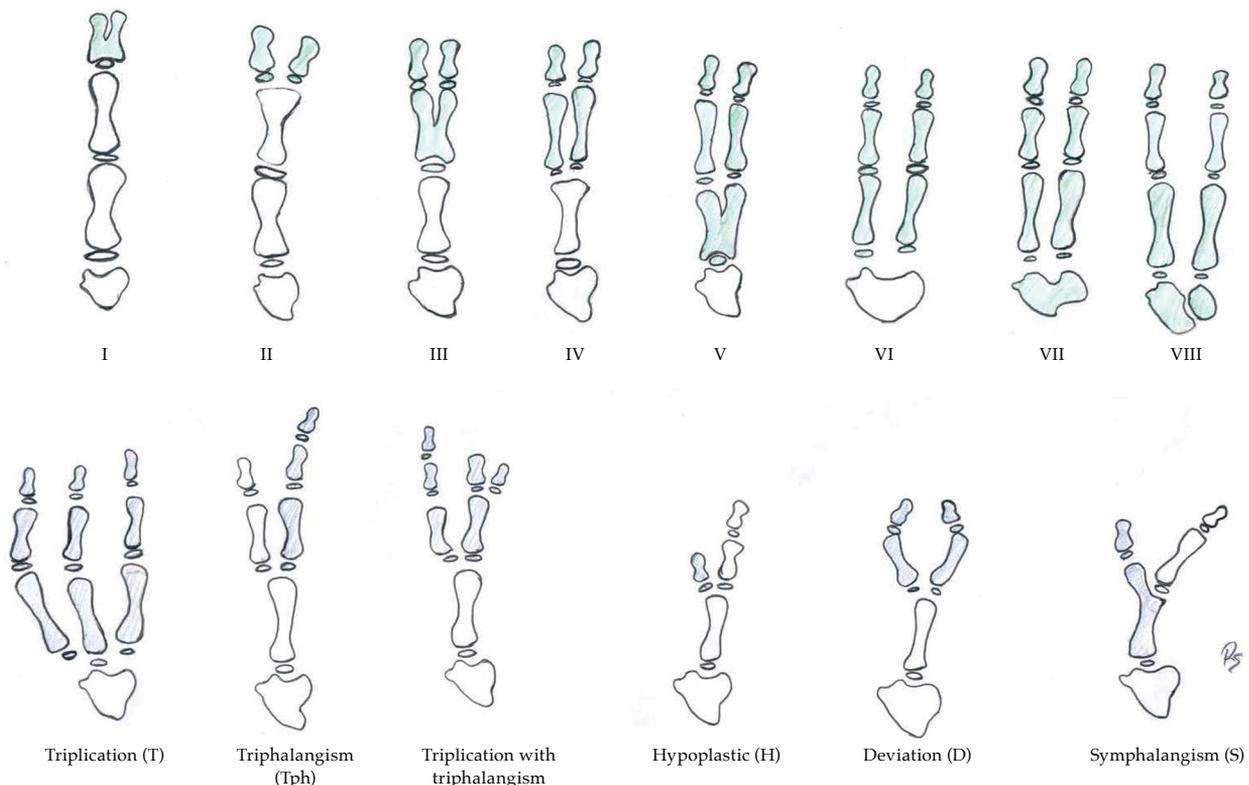


FIGURE 1. Wassel (upper row) and Rotterdam (lower row) classifications.

TABLE I								
Number of patients according to Wassel and Rotterdam classifications								
Classification	I	II	III	IV	V	VI	VII	Unclassifiable
Wassel	Null	5	4	26	1	4	6	5
Rotterdam	Null	7*	4	30†	1	5‡	1	3

* Two cases had a triphalangeal component; † Four cases had a triphalangeal component; ‡ One case had a triphalangeal component.

and evaluated by two of the hand surgeons by consensus, who were trained about Wassel and Rotterdam classification systems (Figure 1). Hand surgeons assessed the cases in random order and classified them according to Wassel and Rotterdam classification systems consecutively.

An intra-class correlation coefficient was calculated to test for an agreement between the two observers for radiographic or anatomic categorization of patients according to the classifications system.

Statistical analysis

Demographic and other descriptive data of our patients were analyzed using the software SPSS 19.0 for Windows (SPSS Inc., Chicago, Illinois, USA) at a significance level of 0.05.

RESULTS

Thumb duplication was present in the right hand in 28 patients, in the left hand in 21 patients, and in both hands in one patient.

Table I shows the distribution of the number of cases according to the classification systems. In our case series, five patients could not be categorized according to the Wassel classification and three

patients could not be categorized according to Rotterdam classification (Figures 2-4). Two cases that were unclassified according to the Wassel classification were identified as type II according to Rotterdam classification, four Wassel type VII cases were found to be Rotterdam type IV, and one Wassel type VII case was identified as type VI in the Rotterdam classification. Wassel type III and V cases were categorized as the same types in Rotterdam classification.

The details of the unclassified cases are presented below:

Case 1- An 11-month-old boy had four thumbs, four other fingers, and a radially deviated wrist. Radiography revealed two ulnae. The lateral-side ulna was distally bifurcated and shorter than the medial one. There were four well-formed ulnar fingers; however, the thumbs did not have the features of the mirror hand. Tetraplication with a hypoplastic ulnar thumb, triphalangeal middle two thumbs, and a hypoplastic radial thumb was detected by radiography. The carpal bones were not visible initially; however, computed tomography of the wrist at the age of seven showed single trapezoid, single lunate, duplicated trapezium, and duplicated scaphoid, unlike ulnar dimelia cases (Figure 2a-d). We could define this case as type X Te, VIu/m, IVr,

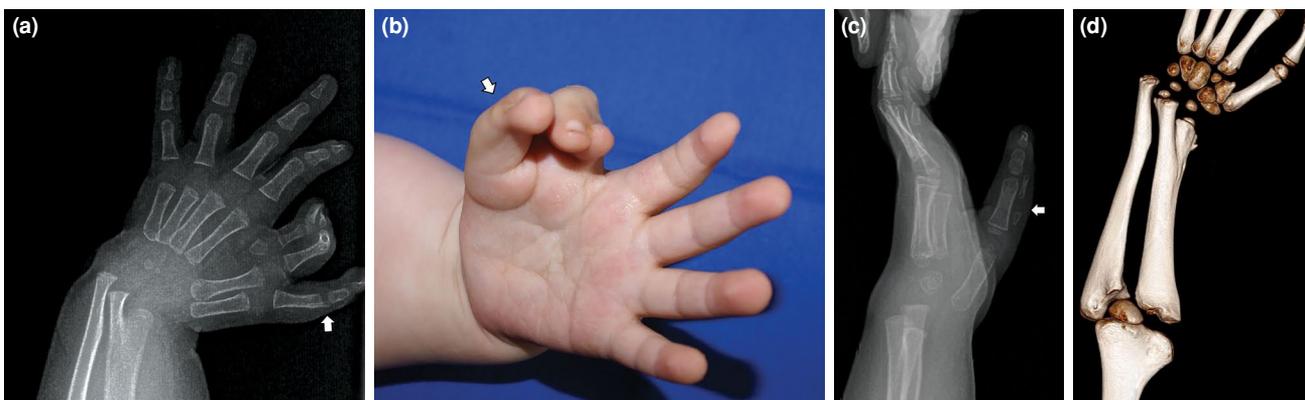


FIGURE 2. (a) Radiograph of the posteroanterior hand and wrist and (b) image of the left hand and wrist at age 11 months. (c) Posteroanterior radiograph of the thumb after the amputation of two ulnar-most thumbs. (d) Three-dimensional computed tomography image of the left hand, wrist, and forearm including the elbow at 7 years of age (white arrowhead denotes the radial-most thumb).

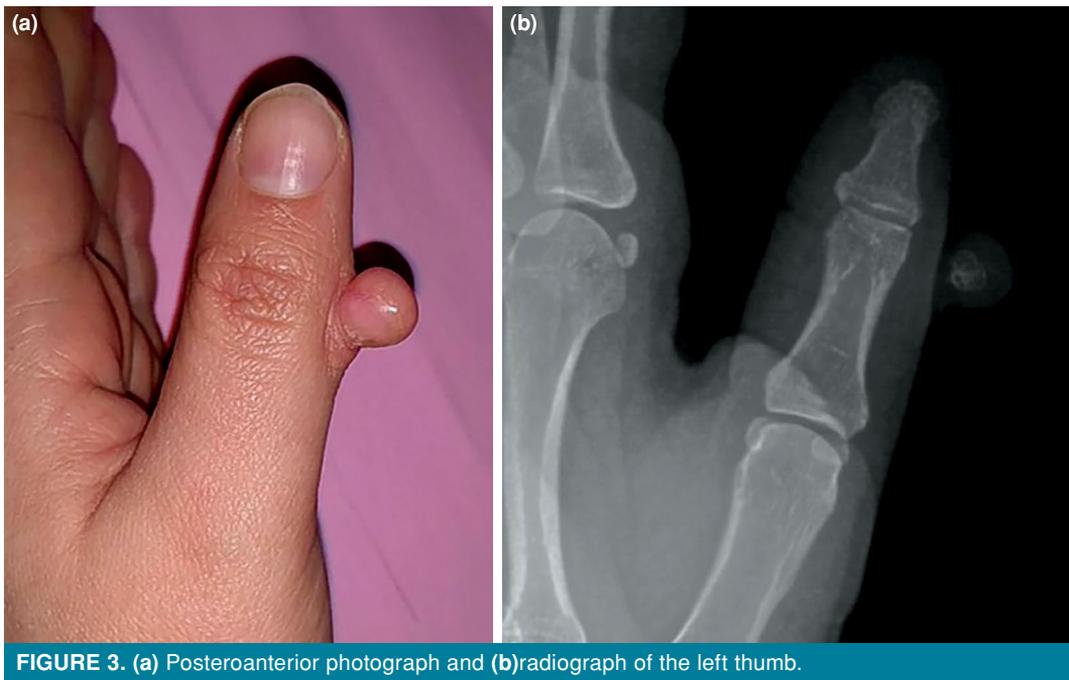


FIGURE 3. (a) Posteroanterior photograph and (b) radiograph of the left thumb.

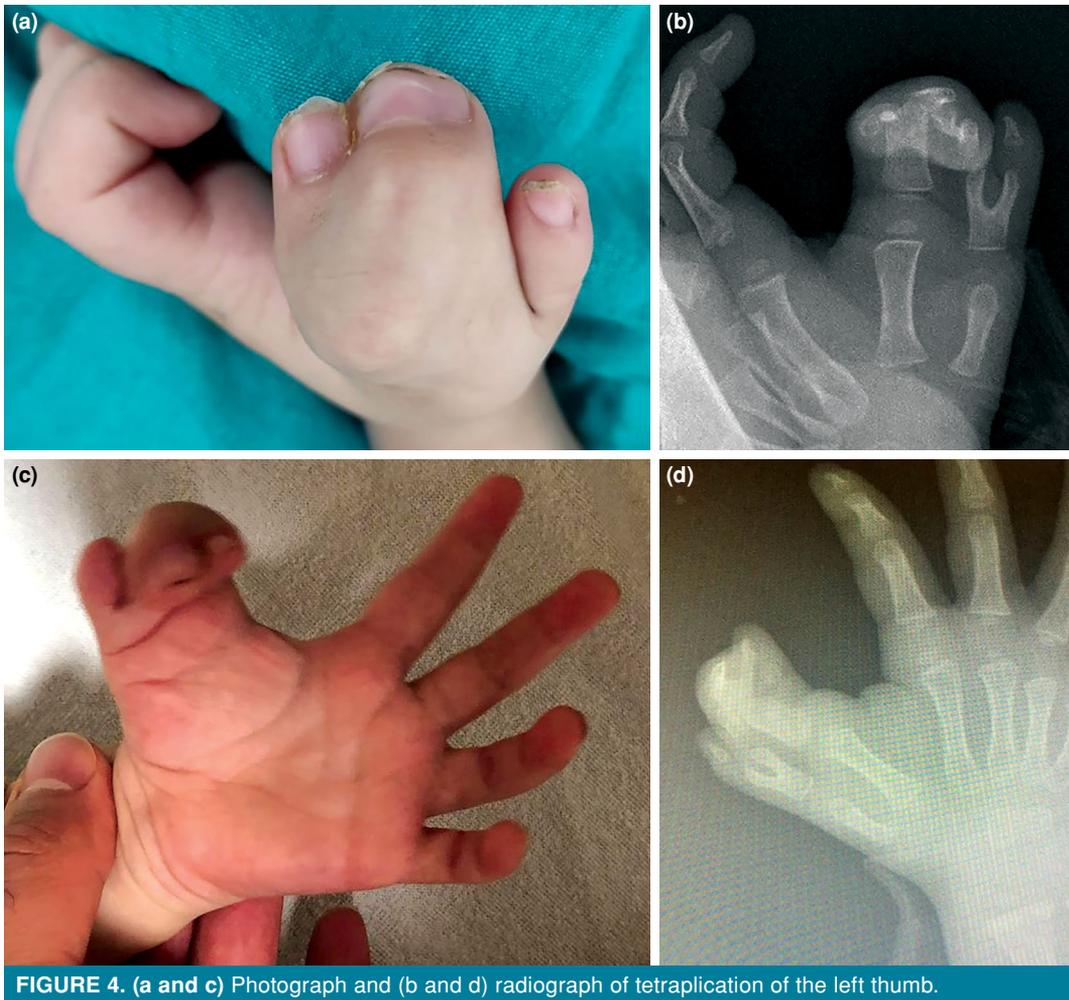


FIGURE 4. (a and c) Photograph and (b and d) radiograph of tetraplication of the left thumb.

Tph2/3, and H1/4 depending on the proposal by Jalili and Najd Mazhar;^[4] however, as this patient had two ulnae, the most proximal level of duplication might require a new definition.

Case 2- A 26-year-old female patient had a small pedunculated structure with a well-formed nail on the radial aspect of a normal looking thumb was observed. Radiograph revealed a small circular bone underlying the nail. There was no bony attachment to the thumb (Figure 3a, b). As the proximal level of duplication was unclear, this case could not be classified in either system.

Case 3- An 18-month-old boy had four thumbs, and three ulnar thumbs were syndactylized. Radiography revealed two metacarpal bones, and the radial one was hypoplastic. The two radial thumbs were similar to Wassel type III duplication, the two ulnar thumbs were duplicated at the interphalangeal level, and the lateral one was triphalangeal (Figure 4a-d). If we attempt to classify this case using the proposal of Jalili and Najd Mazhar,^[4] it would be VI Te, IIu, IIIr Tph3, and syndactylized 2/3/4.

Radiographic categorizations of the patients by two hand surgeons, in both classification systems, were correlated significantly. The values for the correlation coefficient between the two different classification systems were 0.96 for Wassel classification system and 0.99 for Rotterdam classification system.

DISCUSSION

Several studies have attempted to validate the reliability and prognostic value in predicting complications and functional outcome of the classification systems.^[14-16] Nevertheless, a perfect classification system that embraces the whole purposes of hand surgeons dealing with thumb polydactyly have not been developed yet. We described a total of eight cases that were not categorized in at least one of the classification systems previously described in the literature, and three patients that were not categorized in any way according to the previous classifications, together with their radiological and clinical details, in our article.^[17] The classification articles provide guidance in guiding treatment, as well as helping other surgeons to present their cases in an understandable way. Many authors have attempted achieve the best classification;^[6,8] however, these may not include all radial duplication cases, as we have shown in our study. Most triphalangeal, deviating, or hypoplastic cases could not be categorized as any types of

Wassel classification.^[8,16,18] Hu et al.^[16] could not classify 13% of thumbs by Wassel classification. This rate is 9.8% in our study. Rotterdam classification has covered almost all these duplicated cases up to the trapezium. Nevertheless, some authors have pointed out the deficiencies based on their case experiences.^[4,12,13] Triplication of the thumb is a very rare anomaly, and real triplication cases require three different radial rays. Other rarer anomalies may include four thumbs. Our aforementioned Cases 1 and 3 also have four thumbs, which are again very rare deformities and are not mentioned by none of the classifications that we discuss in our study. Jalili and Najd Mazhar^[4] defined a case with bifurcated radius and tetraplication and classified their cases as type X Te, IVu, IIu Tph 2/4, and H 1/3.

Duplication types may vary depending on the ethnicity. Rudimentary duplication is very common among the Far-Eastern population, whereas triplication is more common in communities carrying a mutation in chromosome 7q36 locus. Duplication with cartilaginous symphalangism may be related to a mutation that the Asian genetic pool carries the most.^[19] Hu et al.^[16] also proposed adding a hypoplastic subtype to the Wassel classification, which can be defined as any thumbs that do not have a bone articulation to the larger thumb. By the addition of this subtype they suppose that more cases would be classified by the Wassel classification. Kwang Kim et al.^[18] further divided hypoplastic cases as proximal and distal type. We had only one rudimentary case, Case 2, that could not be classified by the Wassel classification, as it did not have a bony articulation and by the Rotterdam classification and it did not have a clear proximal level of duplication.

One of the drawbacks to the Rotterdam classification is the lack of visibility of certain aberrant components on radiography in young patients.^[20] Radiographs are mostly obtained in patients with immature skeletal system, which may lead to inaccurate evaluation regardless of the classification system.^[7,14,20] It may not be possible to differentiate type VII from type VIII until the patient is approximately five years old, when the ossification of the trapezium occurs, and a mismatch may occur according to the perioperative findings.^[7,9] We depended on radiographs and did not consider the perioperative findings, as both Wassel and Rotterdam classifications are based on radiological findings.

We have some limitations in this retrospectively designed study. First, we do not know the percentage

of consanguineous marriages in the parents of our patients, whether there are similar malformations in the parents or siblings, and whether there are accompanying visceral anomalies. Second, the details of the genetic consultations of these patients are unknown. Third, the number of our cases is not high enough to put forward a new classification.

In conclusion, with the addition of our cases that are unclassified in Wassel classification, in Rotterdam classification, and other case reports, we recommend that the classifications may be carried proximally down to the radius and, so, tetraplication cases may be added.

Declaration of conflicting interests

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