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Unclassified Bilateral Thumb Duplication

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Abstract: Polydactyly being most common congenital digital anomaly of the hand and needs special consideration. Surgical treatment is warranted in most of the cases because of cosmetic and functional reasons. In this article, we describe a 5-year-old female child who was having bilateral thumb duplication. The diagnosis was clinical, and it does not fit into any mentioned classification systems in literature. Patient was operated with resection of supernumerary digit with reconstruction procedure for dominant thumb. Patient was discharged with no complications. The parents were satisfied with the cosmetic and functional result. We discuss types of duplication, and literature review for such cases.

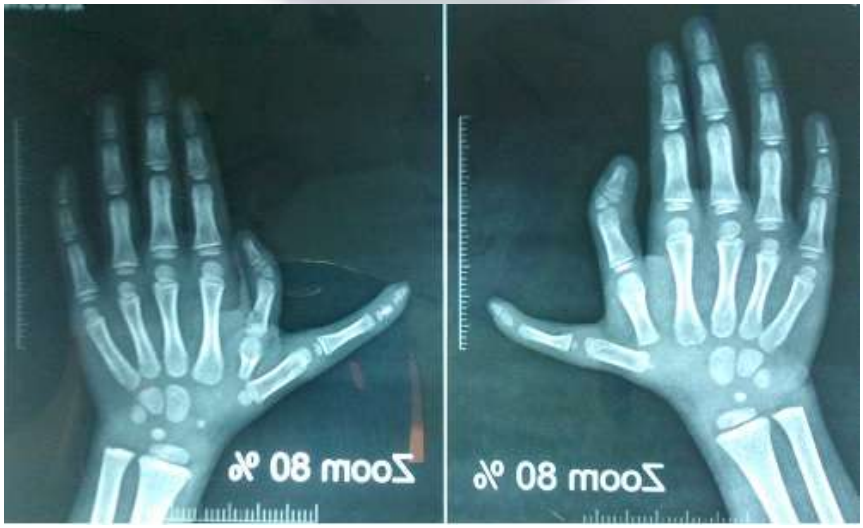
Keywords: Bilateral duplication, Polydactyly, Radial Polydactyly, Supernumerary Digit, Thumb Reconstruction, Thumb duplication.

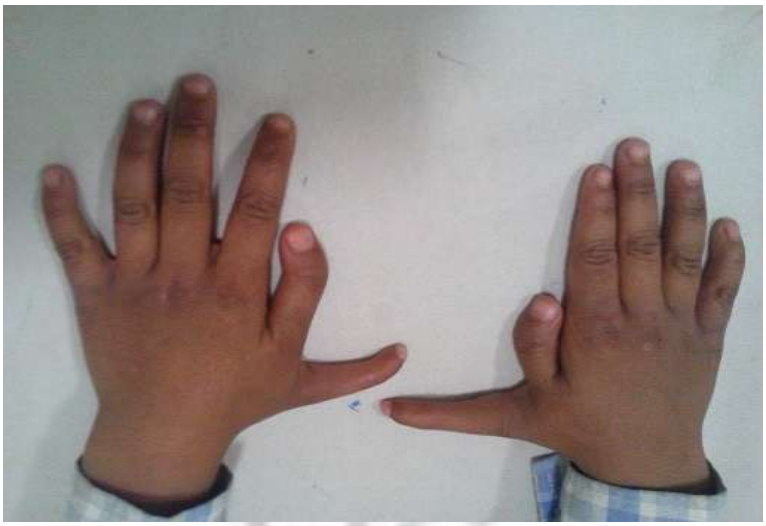
Introduction

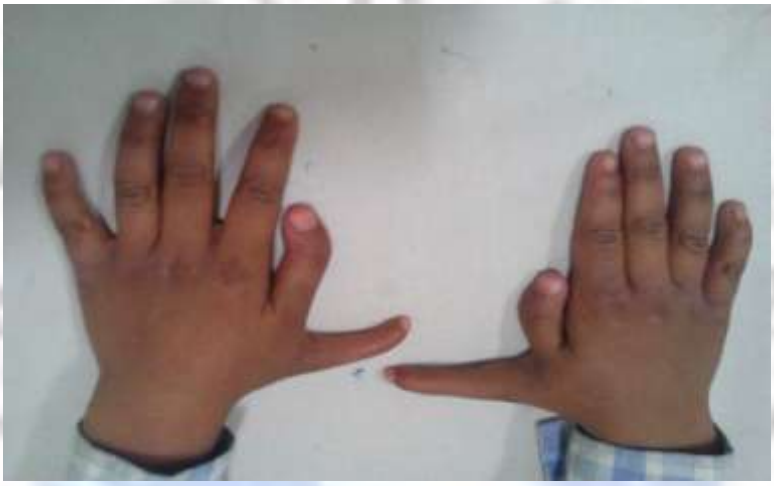
Polydactyly is the most common congenital anomaly of the human hand and has been recorded in literature since 11th century BC[1,2,3]. Thumb duplication is a pre-axial polydactyly and one of the most frequent congenital deformities of the hand after syndactyly. Although it mostly appears an isolated deformity, it can sometimes be associated with other abnormalities[4]. Polydactyly is an abnormality in the longitudinal segmentation of the limb bud which can be classified in literature by different means . No universally accepted classification exist for polydactyly. Preaxial or radial duplication is classified by Wassel[5] into seven types which are further subclassified by Ezaki into types depending upon opposability. Various adaptations and modifications have been added to this scheme, but the basic premise remains unchanged. In addition, there are some types of thumb duplication that defy classification [6,7,8,9] Treatment of each case is different and therapeutic approach depend upon the classification. We report a rare presentation of duplication of thumb which does not fit into any standard classifications mentioned in literature.

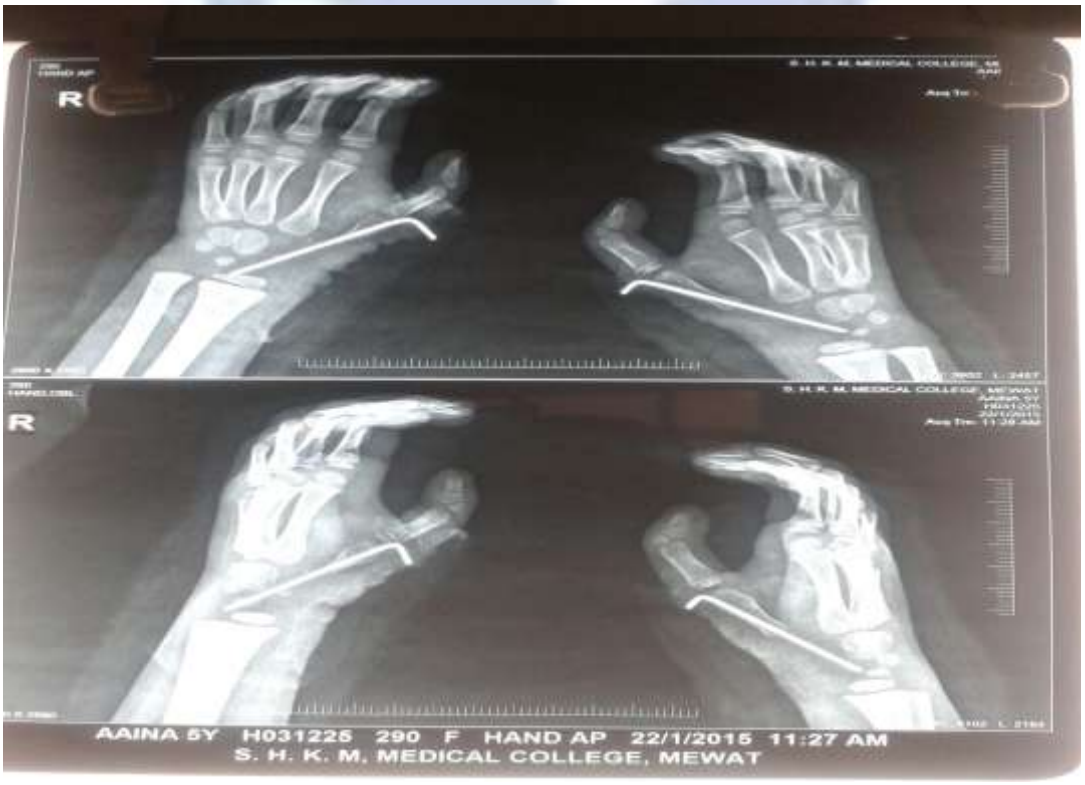
Case Report

5 year female child presented in outpatient department with right hand dominace having bilateral duplication of thumb that was present since birth[Fig-1,2,3,4].





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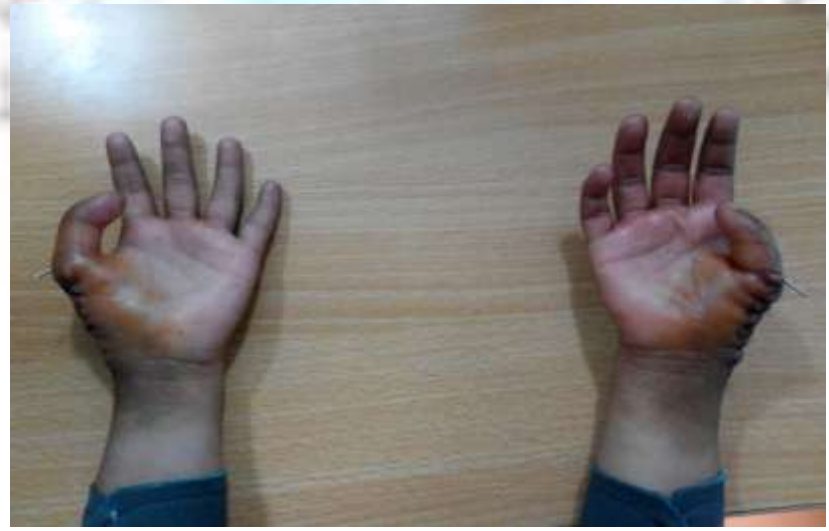
On examination it was preaxial radial duplication of thumbs bilaterally. The Thumbs closer to index finger (Ulnar) on both side were opposable with maximum functions while the radial duplicates were hypopastic without active function. Normal functions of both hands were influenced and disrupted. Standard radiographs of hands showed not exaxct but near configuration of type VII B Wassel with abnormal partial metacarpal of left opposable dominant functional thumb. As such the presentation of this case does not relates to any classification mentioned in literature. Additionally both functional thumbs have middle phalangeal hypoplasia without any functional disruption. Patient though presented late was operated for bilateral excision of supernumerary thumb from base with tendon transfer and restoration of alignment with non absorbable wire for 6 weeks[Fig-4,5,6] . Post operative period was uneventful and patient regained all movements after K wire removal. The joint was stable with satisfactory results.



Discussion

Radial polydactyly (Thumb Duplication) is relatively common and deserves special consideration because the treatment of thumb duplication is so critical and reconstruction is more complex than reconstruction of the typical ulnar polydactyly[10]. Therefore, for useful treatment reasons, hand surgeons have further subclassified these patients. Thumb duplication is a common variety included in preaxial polydactyly. Classification system mentioned in literature for congenital deformities of upper extremities and thumb duplication are:

a] International Classification Type III [figure-7]
b] Stelling and Turek
c] Wassel Classification.[7]
d] Temtamy and McKusick
e] Ezaki classification.

Table 1. International classification of congenital deformities of the upper extremity

Type	Description			Example
I	Formation defects	Transverse	Terminal	Phalangeal, carpal, metacarpal, forearm, upper arm
			Intercalary	Symbrachydactyly, phocomelia
		Longitudinal		Radial (preaxial) or ulnar (postaxial) clubhand, split hand
II	Differentiation (separation)defects			
III	Duplication			Polydactyly, triphalangeal thumb
IV	Overgrowth			Macrodactyly
V	Hypoplasia			Thumb hypoplasia, Madelung deformity
VI	Ring constriction syndrome			–
VII	Generalized skeletal anomalies			Apert syndrome, Poland syndrome, arthrogryposis

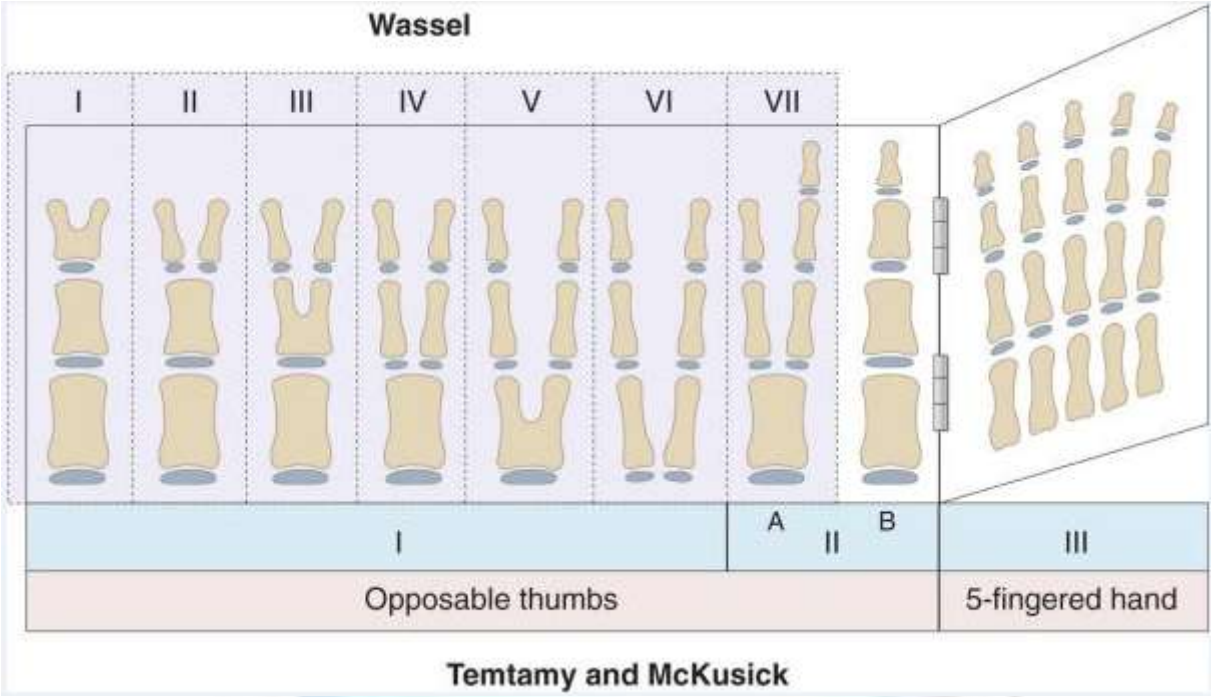
Internationally accepted and most common age to operate for various congenital conditions are described in figure -8. In our case the presentation of the child was unique and reporting to outpatient clinic was delayed but the post surgery results remains satisfactory.

Table 2. Most favorable ages for surgery

Deformity	Condition/Operation	Age
Syndactyly	Simple	12 months
	Bony	8 months
	Acrosyndactyly	4 months
Clubhand	Centralization	12 months
	Pollicization	2 years
	Lengthening	12 years
Polydactyly	5th finger	4 months
	Thumb	1 year
Finger aplasia	Pollicization	1–2 years
	Finger transfer	1–2 years
	Lengthening	12 years
Ring constriction syndrome	With vascular impairment	Emergency
Symbrachydactyly	Finger stabilization	1–2 years
Delta phalanx	Osteotomy	3–4 years
Radioulnar synostosis	Osteotomy	7–8 years

The surgical repair of radial polydactyly is usually more complex than the surgical repair of ulnar duplications. "Thumb reconstruction" describes this procedure well because ligamentous reconstruction, angular correction, and tendon reconstruction are usually required to make the best of this most important digit. Although the thumbs may be symmetric or asymmetric, the tendons usually bifurcate from a normal proximal tendon. The tendon insertions of both flexor and extensor are frequently eccentrically placed and must be repositioned and aligned by the reconstruction[10]. The case we reported have a unique pattern of thumb duplication which does not fit into any available classification systems. Right

sided duplicated thumb anomaly showed complete duplication with additional hypoplastic phalanx while left sided duplicated thumb have hypoplastic metacarpal with additional hypoplastic phalanx.. Therefore, for treatment purposes hand surgeons have subclassified these patients. Wassel developed seven types [figure-9]and reported their frequency:



Type I 2%
Type II 15%
Type III 6%
Type IV 43%
Type V 10%
Type VI 4%
Type VII 20%.

The geneticists Temtamy and McKusick divided the preaxial polydactylies into three groups. This classification is particularly useful clinically because of the correlation of the triphalangeal thumbs with and without duplication and the true five-fingered hand. Temtamy and McCusick's subclassified as:

Type I - Radial polydactyly includes the first six types of Wassel.
Type II- Radial polydactyly includes two varieties of opposable thumbs:
IIA- duplicated thumbs with a triphalangeal member (Wassel's type VII);
IIB- nonduplicated triphalangeal thumbs.
Type III- Radial polydactyly includes nonopposable duplication of the index finger, also known as a five-fingered hand.

Conclusion

In thumb duplication, neither of the two thumbs is equal in size to a normal thumb, even when they are grossly different from each other. Before Surgery parents should be well explained about outcome of procedure. In-spite of numerous available classification systems, presentation of any kind is feasible which indicate that a lot of work is still need to be done in cases of bilateral thumb duplication. Simple excision is seldom indicated now-a-days. Various surgical techniques can be combined involving procedures of bone, soft tissues, joint and tendons are preferred for reconstruction.The major current aim should be to achieve the maximal good result with a minimal number of surgical sittings.The Surgical procedure to be done for a particular case needs to be tailored after thorough clinical and radio-logical study.

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