

Thumb polydactyly: clinical outcome after reconstruction

CH Yen, WL Chan, HB Leung, KH Mak

Department of Orthopaedics and Traumatology, Kwong Wah Hospital, Hong Kong

ABSTRACT

Purpose. To evaluate clinical and cosmetic outcomes of reconstruction in thumb polydactyly and prognostic value of the Wassel classification.

Methods. Between 1993 and 2000 inclusive, out of the patients with thumb polydactyly (involving 80 thumbs) operated on, 34 patients (36 thumbs) were available for review and underwent clinical and radiological assessment. Outcomes in terms of the Tada score and complications were recorded.

Results. The mean age of patients at the time of operation was 2.8 (range, 0.6–47) years. The mean follow-up period was 5 (range, 2.4–10) years. According to the Wassel classification, 12 were type-II thumb polydactyly, 3 type-III, 11 type-IV, 6 type-V, one type-VI, and 3 type-VII. There was no perioperative mortality or wound infection. More than 88% of the patients were satisfied or very satisfied with functional and cosmetic outcomes. Postoperative complications such as scar hypertrophy, pulp atrophy, joint deformity, and instability were common but minor. Ridge nail deformity after the Bilhaut Cloquet procedure was amenable to second-

ary corrective procedures. All types of operated thumb polydactyly achieved similar mean Tada scores (14.7–16.6 out of 20). The Wassel classification category, age, and surgical procedures were found to have no prognostic value with regard to the Tada score and presence of complications.

Conclusion. Surgery on thumb polydactyly is rewarding. The Wassel classification category can be used as a guide for treatment, although it fails to predict the occurrence of postoperative complications or Tada scores. Our patients' results can serve as guidelines of expected outcomes after reconstructive procedures in different sub-types of thumb polydactyly.

Key words: polydactyly; thumb; treatment outcome

INTRODUCTION

Thumb polydactyly is the most common type of polydactyly in the hand. It is believed to arise from excessive cell proliferation and disturbed cell necrosis of pre-axial ectodermal and mesodermal tissues before the eighth week of embryonic life.¹ It occurs sporadically with an incidence of 8 in 100 000 in both black and white populations.^{2,3} Hereditary

influence has not been documented in isolated thumb polydactyly, although Ezaki² found its association in several syndromes. Autosomal dominance has been reported only in triphalangism and polysyndactyly.

The psychological burden of having an extra finger has been linked with psychosis in adulthood.⁴ Operation remains the definitive treatment with a goal to improve cosmesis and possibly hand function.⁵⁻⁷ Since Bilhaut first described an operation for thumb polydactyly in 1890, different surgical procedures based on the type of radiographic abnormality have been reported.⁸⁻¹⁰ Currently, the Wassel classification is universally accepted to categorise the patho-anatomy of the polydactyly and to guide respective surgical procedures.⁵ However, its prognostic significance has been speculative as long-term outcomes of surgical treatment are seldom reported.¹¹⁻¹³

We aimed at evaluating surgical outcomes and complications according to the Wassel classification category and its prognostic value, particularly to address the effectiveness of secondary surgical procedures to rectify residual deformities, ridge nail deformity, and Z thumb deformity.

MATERIALS AND METHODS

Polydactyly affecting 80 thumbs were operated between 1993 and 2000 in Kwong Wah Hospital. A dedicated surgical team performed all operations according to the guidelines on thumb reconstruction (Table 1). All patients were allowed immediate postoperative free mobilisation of all digits in bulky dressings. 34 of them (entailing 36 polydactyly thumbs) who had complete medical records and pre-operative radiography were available for assessment. Intra-operative findings of soft tissue and osseous anomalies and details of the respective surgical procedures were retrieved from the medical record.

Review and measurements were conducted by one of the authors, blinded to the type of polydactyly (to avoid bias). Postoperative functional outcome was evaluated using the Tada score¹⁴ comprising of 3 evaluations: cosmetic, functional, and radiological. Patients were assessed clinically with respect to nail width of the operated thumb and cosmetic acceptability. Grip strength, tip and chuck pinch were measured using a Jamar Dynamometer (JA Preston Corp, Jackson [MI], USA) and pinch gauge (B&L Engineering, Pinsco Inc, Santa Fe Springs [CA], USA). Assessment specifically addressed first web contracture and bone growth. Evaluation of growth potential, instability, and deformity were performed

Table 1
Guidelines for operating on thumb polydactyly

1. Hypoplastic radial thumbs are usually resected.
2. In types-II and -III thumb polydactyly with symmetrical calibre and osseous components, the Bilhaut Cloquet procedure is preferred.⁶
3. Radial collateral ligaments with their attached periosteal flaps are preserved and over-tightened to confer joint stability and prevent deformity.
4. Types-II and -IV thumbs are usually associated with an excessively large proximal phalanx and metacarpal head, respectively. Chondroplasty on the radial facet is usually performed to trim down the size of the proximal articulating surface in order to restore joint stability, mobility, and axial alignment.
5. Corrective osteotomy is preferred for any residual bony angular deformity.
6. Re-alignment with or without augmentation of the tendon is important to restore axial alignment and prevent Z deformity due to eccentric tendon pull. In type IV, the usual procedure includes suturing of duplicated extensor of radial digit to the long extensor of the ulnar digit and reattachment of abductor pollicis brevis and extensor pollicis brevis to the base of proximal phalanx.

clinically and radiologically. X-rays of thumbs were taken in posteroanterior view in full extension, stressed radially and ulnarly (without undue force), with the opposite thumb as self-reference. Full marks on the Tada score was 20.

Definition of growth parameters, first web contracture, stiffness, deformity, and instability were based on the Tada score. Retarded radial growth was defined as the thumb circumference of the distal and/or proximal phalanx being <75% of that on the normal side. Longitudinal growth was represented by the longitudinal length of the thumb between the carpometacarpal joint (CMCJ) and the tip of thumb in the resting position. Measuring the width of nail plate quantified nail growth. The terms 'hypertrophic scar' and 'atrophic pulp' were based on the perceptions of the patients or their guardians. First web contracture was defined as a loss of >25% of intermetacarpal angle between the first and second metacarpal bones, when compared to the normal side. Stiffness was defined as a loss of >25% of the sum of active interphalangeal joint (IPJ) and the metacarpophalangeal joint (MCPJ) range of movement with reference to the normal side. Ulnar deformity was defined as >5° of ulnar angulation at the IPJ or >20° of angulation at the MCPJ in the resting position. Radial instability was defined as >5° of angulation at the IPJ

or $>20^\circ$ at the MCPJ when the examiner subjected the thumb to maximal stress from the resting position. In cases of bilateral polydactyly, the digit with the less affected parameter was taken as the normal side for comparison.

Data were summarised and tabulated. Prognostic factors for the presence of complications were identified using a backward binary logistic regression model. The relationship of the Tada score to age was subjected to correlation analysis. Mean Tada scores were compared between different types (based on the Wassel classification category) by analysis of variance. Data analysis was performed using the Statistical Package for the Social Sciences (Windows version 9.0; SPSS Inc, Chicago [IL], USA).

RESULTS

The case series comprised 24 male and 10 female patients; 2 males had bilateral, asymmetrical involvement; 16 left and 20 right hands were affected. The distribution ratio of different types of thumb polydactyly closely resembled previous reports.¹³⁻¹⁵ The mean age of the patients at the time of operation was 2.8 (range, 0.6-47) years and the mean follow-up period was 5.3 (range, 2-10) years (Table 2).

Three male patients had a family history of thumb polydactyly in first-degree relatives (all being mother). Another male patient had a positive family history of thumb polydactyly in his paternal grand-grandfather. Three had associated spinal or renal abnormalities. One was diagnosed to have the CHARGE syndrome (Coloboma of the eye, Heart defects, Atresia of the choanae, Retardation of growth and/or development, Genital and/or urinary abnormalities, and Ear abnormalities and deafness) with choanal atresia and respiratory distress.

The mean nail width of the affected thumb was 1.4 (range, 0-4) mm narrower than that of the normal side. In 2 cases of bilateral polydactyly, the digit with the wider nail was taken as the control. All 6 cases of type-III and -VII thumbs had normal radial growth, whereas 2 of the 12 type-II, 2 of the 11 type-IV, one of the 6 type-V, and one type-VI thumbs had smaller thumbs in terms of circumference. As to longitudinal growth, all patients had equal thumb lengths regardless of the type of polydactyly.

Postoperative complications are tabulated in Table 2, including pulp atrophy (12 digits), scar hypertrophy (8 digits), joint instability (25 digits), joint deformity (10 digits), web space contracture (4 digits), and joint stiffness (8 digits). With back-

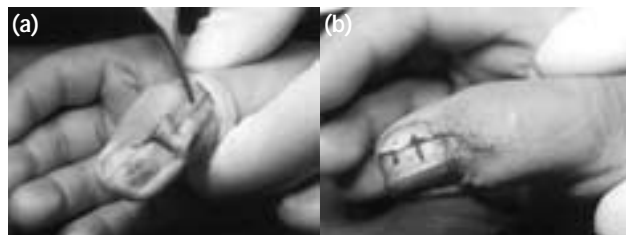


Figure 1 Ridge nail deformity: (a) scarring at apposed edges of nail bed, (b) replacement of nail plate after excision of scarred tissue and meticulous repair of nail bed.

ward binary logistic regression, no prognostic factor (including Wassel type, gender or age) was identified for any of these complications.

Four patients were found to have postoperative first web contracture, but there was no functional impairment. Six and 24 patients, respectively, were very satisfied and satisfied with their operations; only 4 were dissatisfied with the cosmetic outcome.

Three thumbs were complicated by over-sized nail with ridge nail deformities; all had undergone the Billhaut Cloquet procedure.⁶ One patient was dissatisfied with the cosmetic outcome and underwent a secondary rectification procedure. The nail plate was elevated to excise scarred tissue along the ridge in the nail bed; paronychia was then reconstructed (Fig. 1). The pathology of excised scarred tissue revealed fibrosis of the nail bed in the absence of keratinisation. At the latest follow-up, the nail plate had resumed normal growth without residual deformity.

A case of Z thumb deformity was observed in a type-III polydactyly. The patient had undergone the first operation elsewhere and presented to our department 4 years later. The exact nature of that operation was unknown but Z thumb deformity was present; there was a tight capsule and scarring in the radial collateral ligament with laxity in the ulnar capsule at the IPJ, angular deformity of the proximal phalanx, and an eccentric pull of the malaligned extensor pollicis longus (EPL). Release of scarred tissue on the radial side, capsulorrhaphy on the ulnar side, corrective osteotomy of the proximal phalanx with wire loop fixation and re-alignment of the EPL were performed, which achieved a satisfactory cosmetic and functional recovery (Fig. 2).

Analysis of variance did not detect a significant difference in Tada scores between different Wassel types of thumb polydactyly. Age was not related to the Tada score, according to the correlation analysis.

Table 2
Clinical results of treating various types of polydactyly

Type	Gender	Age at operation (months)	Procedures						
			Excision of hypoplastic digit	Bilhaut Cloquet procedure	On-top plasty	Chondroplasty	Osteotomy	Collateral/capsular reattachment	EPL/APL/APB/ABD/ADD* reattachment
II	F	15	✓					✓	
II	M	24	✓			✓		✓	
II	M	35	✓					✓	
II	M	20	✓					✓	
II	F	28	✓					✓	
II	M	13		✓					
II	F	18	✓						
II	M	73	✓						✓
II	F	26	✓						
II	M	226	✓						
II	F	10		✓					
II	M	10	✓						
Mean		41.5							
III	F	20		✓			✓		
III	M	12	✓						
III	F	19	✓					✓	✓
Mean		17							
IV	M	12	✓						
IV	M	66	✓				✓		
IV	F	7	✓						
IV	M	28			✓				
IV	M	13	✓					✓	✓
IV	M	19	✓						✓
IV	F	564	✓						
IV	M	28	✓						
IV	M	9	✓						
IV	M	19	✓						
IV	M	26	✓						
Mean		71.9							
V	M	28	✓				✓		✓
V	M	19	✓					✓	✓
V	F	27	✓						
V	M	28	✓						✓
V	M	19	✓						✓
V	M	11	✓						
Mean		22							
VI	M	24	✓						✓
Mean		24							
VII	M	23	✓						✓
VII	M	23	✓						✓
VII	M	13	✓				✓		✓
Mean		19.7							

* EPL denotes extensor pollicis longus, APL abductor pollicis longus, APB abductor pollicis brevis, ABD abductor, and ADD adductor

DISCUSSION

Despite minor complications from reconstruction of thumb polydactyly, the operated thumbs are cosmetically and functionally acceptable, provided guide-

lines on reconstruction are strictly observed. In our series, more than 88% of patients were satisfied or very satisfied with the surgical procedure. Thus the surgical outcome appeared to have met patient and parent expectations; their prime concern being removal

Extensor/ flexor augmenta- tion	Z-plasty	Follow-up (years)	Tada score	Postoperative complications					
				Pulp atrophy	Scar hyper- trophy	Joint instability	Joint deformity	Web space contracture	Joint stiffness
		9	17	✓		✓			
		4	17						
		5	16			✓			
		2	14			✓			
		4	15	✓		✓			
		9	19						
		4	15			✓		✓	
✓	✓	6	15			✓			✓
		6	17	✓			✓		
		7	20						
		7	15	✓	✓				✓
		5	19						
		5.7	16.6						
		8	15			✓			
		5	15		✓	✓			
		10	14	✓	✓	✓			✓
		7.7	14.7						
		5	14			✓		✓	
		5	19	✓					
✓		7	15		✓	✓			
		3	18	✓					
		5	16	✓					✓
		3	12	✓		✓	✓		
		3	18			✓			
		4	18			✓			
		4	14			✓	✓		
		8	12			✓			
		3	10	✓	✓	✓	✓		✓
		4.5	15.1						
		4	11			✓	✓		
		4	16			✓		✓	
✓		5	16	✓	✓	✓	✓		
		4	15		✓	✓			✓
		9	17			✓	✓		
		4	16		✓	✓			✓
		5	15.2						
		4	15			✓			
		4	15						
		4	15			✓	✓	✓	✓
		4	15						
		7	15	✓		✓	✓		
		5	15						

of the supernumerary digit. The Wassel classification was used for its simplicity for the purposes of clinical categorisation and planning of surgical procedures.

For type-II polydactyly, joint instability is quite common as the deformity develops at joint level.

Collateral ligaments, capsular attachment, and the extrinsic tendon from the hypoplastic thumb are essential structures to preserve, in order to achieve joint stability. Despite following all the guidelines on thumb reconstruction, 6 (50%) of these 12 patients

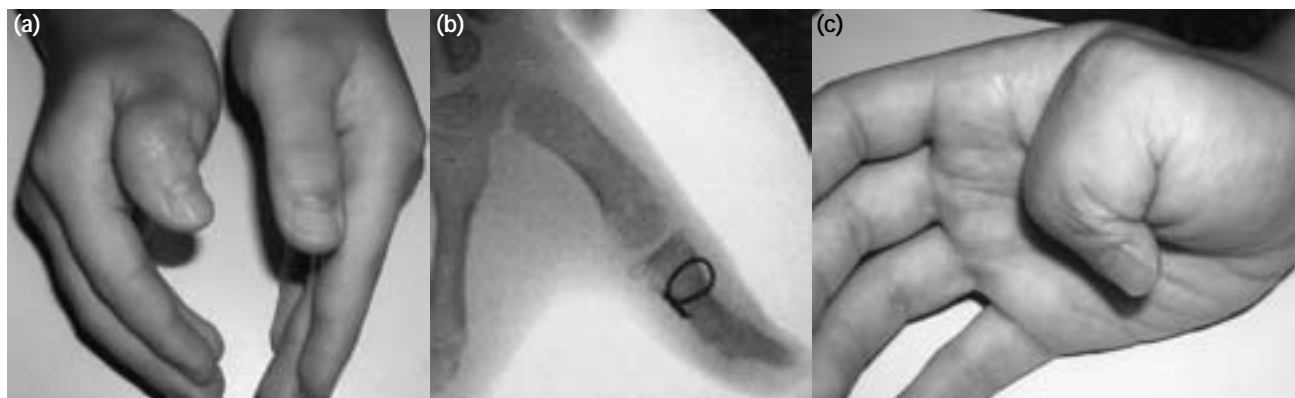


Figure 2 Z thumb deformity in type-III polydactyly: (a) tight capsule and scarring in radial collateral ligament at interphalangeal joint, laxity in the ulnar capsule, angular deformity of proximal phalanx, and eccentric pull of malaligned extensor pollicis longus (EPL) drawing the distal phalanx. (b) Radiographic result after release of scarring tissue on radial side, capsulorrhaphy on ulnar side, osteotomy of proximal phalanx with wire loop fixation, and re-alignment of EPL. (c) Good cosmetic and functional results after secondary procedures.

ended up with joint instability. Late instability may result from soft tissue yielding to chronic stretching or unbalanced soft tissue reconstruction. Therefore, over-tensioning of these soft tissue reconstructions is preferable. However, assessment of joint instability ($>5^\circ$ of angulations at IPJ) is invariably subject to intra-observer errors, particularly if the patients are very young. Nonetheless, 2 of our patients who underwent the Bilhaut Cloquet procedures ended up with stable joints, as it preserves both radial and ulnar collateral ligaments of the IPJ. Complications following this procedure include: joint stiffness, scar hypertrophy, and ridge nail deformity.¹² Meticulous repair of the nail bed and reconstruction of a similar nail size are necessary to prevent these disfiguring problems. It is also important to warn patients of the inevitable hypoplasia of the remaining digit, in terms of nail width and thumb circumference.

For type-III polydactyly, the anomaly does not breach the IPJ and a good functional result is expected after simple excision. However, all 3 patients had unstable IPJs postoperatively. Though not directly arising from the site of the anomaly, one of them had a Z thumb deformity at the IPJ, distal to the site of polydactyly. Five of the 6 type-V polydactyly patients also had joint instability and 3 had joint deformity. Lourie et al.¹⁶ also reported Z thumb deformity after simple suture ligation of the radial component of a bifid thumb. As preoperative joint stability was not documented in our study, joint instability might have been due to an inherent defect. Eccentric pull of extensors across the IPJ might be contributory,

leading to secondary changes in the joint capsule and collateral ligaments. Over-tightening of collateral ligaments and proper re-alignment of extrinsic tendons can correct joint instability. In terms of joint stability, the Bilhaut Cloquet procedure does not appear to offer additional benefit over simple excision in type-III polydactyly, which is in contrast to what ensues in the type-II polydactyly. Despite minor joint instability, all type-III patients were satisfied with their functional and cosmetic outcomes.

In type-IV polydactyly, 8 of the 11 patients had joint instability, 3 also had joint deformity. These complications are more conspicuous in view of the larger size of the MCPJ and proximally based deformity. One of 4 patients who underwent resection of the oversized, bifaceted first metacarpal head followed by MCPJ reconstruction developed joint stiffness. The abnormality in size and contour of articular surface of the metacarpal head was presumably the culprit. These complications can be prevented by careful chondroplasty, using a sharp scalpel to construct a matching articular surface on the base of the proximal phalanx. The outcome of on-top plasty (transposition of distal part of one digit to proximal part of another digit) was good, resulting in a stable thumb with normal alignment (Fig. 3). This procedure was applicable in type-IV polydactyly provided that the ulnar digit had an equal calibre and intact functioning tendinous units were attached. When the ulnar digit was transposed to the base of the radial component, the base of proximal phalanx of the ulnar component was removed. The ulnar articular surface

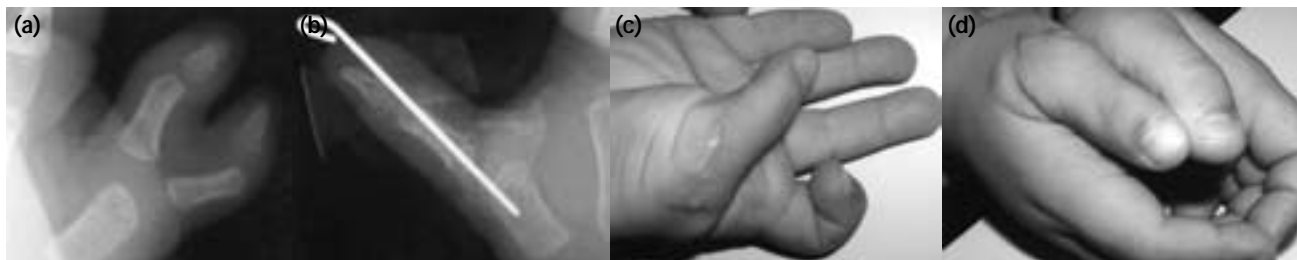


Figure 3 On-top plasty in type-IV polydactyly: (a) preoperative abnormality, (b) transposition of ulnar component to base of radial component with temporary fixation by Kirschner wire, (c) cosmetic and (d) functional results.

of the metacarpal head was trimmed to form a stable base, matching the size of the proximal phalanx of the radial component. This procedure preserved the integrity of the important soft tissue envelope on the radial side, specifically the collateral ligament, the capsule, and the thumb abductor. Intra-osseous Kirschner wire was used to temporarily transfix the osteotomy (Fig. 3). Attention was paid to proper re-alignment of tendons to the new axis of the reconstructed thumb. The procedure provided better bone union and avoided the potential late complications of capsular and collateral reconstruction on the radial side.

Although type-VII polydactyly is morphologically and radiographically distinct according to the Wassel classification, in terms of the Tada score and complications its clinical outcomes did not differ from other subtypes.

Different types of reconstructive procedures gave satisfactory functional and aesthetic surgical outcomes, in spite of pulp atrophy and scar hypertrophy. The ultimate concern of all parents is reconstruction of a functional thumb with ablation of the hypoplastic digit. Despite minor complications from surgery, it is possible to attain a good functional outcome in terms of its length, mobility, strength and stability. Regardless of the Wassel type, our results demonstrate that a longitudinal splitting of the pre-axial anlage does not affect longitudinal growth.¹

As the Wassel classification only categorises the osseous patho-anatomy in radiographs, a variety of intra-operative soft tissue abnormalities associated with polydactyly (eccentric tendons, over-sized articular cartilage, ligamentous and capsular laxity) are not taken into account. Such abnormalities can result in minor complications such as web space contracture, joint deformity, joint stiffness, and joint instability. Intra-operative awareness of these possibilities is therefore of utmost importance. Over-tightening of the lax capsule and ligament and proper

re-alignment of eccentric tendons can prevent joint deformity and instability. Postoperative immobilisation by a short arm brace can protect soft tissue reconstruction, which should be followed by rehabilitation (to prevent joint stiffness). Joint instability, stiffness, and deformity are not necessarily confined to the joint level from which the polydactyly arises. They may result from inherently associated soft tissue anomalies and therefore preoperative and intra-operative assessment become essential. The most disfiguring complications were ridge nail and Z thumb deformities, which were nevertheless amenable to secondary procedures (meticulous nail bed repair and osteotomy, respectively).

The growth potential of the reconstructed thumb is unknown. Measurements of the nail width, thumb circumference, and thumb length reflect the growth potential of the remaining thumb after excision of the hypoplastic digit. In our series, thumb polydactyly hypoplastically affected the nail plate and thumb circumference. Although surgery was usually performed at an early age, nail and circumferential growth of the remaining thumb never caught up with that of the normal thumb. Therefore, the projected growth of a reconstructed thumb is not expected to be normal, even with early removal of the redundant digit. There was no correlation between the Tada score and age, nor did age at surgery affect the growth potential and final outcome of the reconstructed thumb. Whether to perform surgery at an earlier age requires considerations about technical difficulty being balanced against the need to avoid social and psychological stigmatisation. Although more technical difficulty is encountered in younger patients, in this study there was no increase in morbidity.

Although the Wassel classification was not found to affect the Tada score, it was useful to define the osseous part of patho-anatomy and guide decisions for operations. The Tada score is a comprehensive instrument, which has been validated in the assess-

ment of surgical outcomes of thumb polydactyly, as it takes cosmesis, functional, and radiographic evaluations into account.¹⁴ Thumb strength is mostly dependent on joint stability and muscle power. It is difficult to assess grip and pinch strength in very young patients, as they are either too weak to perform the test or too immature to follow instructions. Its significance in clinical documentation in the very young age-group is doubtful. Except for 2 patients (aged 18 and 47 years), the age at operation for the remaining 32 patients were ≤ 7 years. One of our adult patients still had minor joint instability, indicating inherent soft tissue abnormality and was likely to have ensued irrespective of the age at which operation was performed. Despite prevalent minor complications, all patients invariably achieved reasonably high Tada scores. Soft tissue is subject to change with time and growth. Therefore, a longer follow-up (until skeletal maturity is attained) may be more valid to determine the final outcome of surgery.

CONCLUSION

Reconstruction of thumb polydactyly is a rewarding intervention with satisfactory cosmetic and functional outcomes. It is important to use the guidelines of thumb reconstruction, paying particular attention to the soft tissue components. Complications are common but often minor. Those such as nail ridge deformity and Z thumb deformity are amenable by corrective surgery, with promising cosmetic and functional outcomes. The Wassel classification is a useful guide for the treatment of different types of thumb polydactyly.

ACKNOWLEDGEMENT

We wish to express our gratitude towards the Hong Kong College of Orthopaedic Surgeons for its research grant.

REFERENCES

1. Yasuda M. Pathogenesis of preaxial polydactyly of the hand in human embryos. *J Embryol Exp Morphol* 1975;33:745–56.
2. Ezaki M. Radial polydactyly. *Hand Clin* 1990;6:577–88.
3. Graham TJ, Ress AM. Finger polydactyly. *Hand Clin* 1998;14:49–64.
4. Purandare N, Plunkett S. Co-occurrence of polydactyly and psychosis. *Br J Psychiatry* 1999;174:460.
5. Wassel HD. The results of surgery for polydactyly of the thumb. *Clin Orthop Relat Res* 1969;64:175–93.
6. Light TR. Treatment of preaxial polydactyly. *Hand Clin* 1992;8:161–75.
7. Cohen MS. Thumb duplication. *Hand Clin* 1998;14:17–27.
8. Hung L, Cheng JC, Bundoc R, Leung P. Thumb duplication at the metacarpophalangeal joint. Management and a new classification. *Clin Orthop Relat Res* 1996;323:31–41.
9. Manske PR. Treatment of duplicated thumb using a ligamentous/periosteal flap. *J Hand Surg Am* 1989;14:728–33.
10. Masuda T, Sekiguchi J, Komuro Y, Nomura S, Ohmori K. "Face to face": a new method for the treatment of polydactyly of the thumb that maximises the use of available soft tissue. *Scand J Plast Reconstr Surg Hand Surg* 2000;34:79–85.
11. Ganley TJ, Lubahn JD. Radial polydactyly: an outcome study. *Ann Plast Surg* 1995;35:86–9.
12. Ogino T, Ishii S, Takahata S, Kato H. Long-term results of surgical treatment of thumb polydactyly. *J Hand Surg Am* 1996;21:478–86.
13. Goffin D, Gilbert A, Leclercq C. Thumb duplication: surgical treatment and analysis of sequels. *Ann Chir Main Memb Super* 1990;9:119–28.
14. Tada K, Yonenobu K, Tsuyuguchi Y, Kawai H, Egawa T. Duplication of the thumb. A retrospective review of two hundred and thirty-seven cases. *J Bone Joint Surg Am* 1983;65:584–98.
15. Cheng JC, Chan KM, Ma GF, Leung PC. Polydactyly of the thumb: a surgical plan based on ninety-five cases. *J Hand Surg Am* 1984;9:155–64.
16. Lourie GM, Costas BL, Bayne LG. The zig-zag deformity in pre-axial polydactyly. A new cause and its treatment. *J Hand Surg Br* 1995;20:561–4.