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Congenital Clinodactyly of the Thumb

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Author's contribution

This whole work was carried out by author NKS.

Case Study

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ABSTRACT

Aims: To present five patients with thumb clinodactyly and the results of surgical treatment in two patients who were followed till skeletal maturity.
Case Presentation: Four patients (five thumbs) exhibited an ulnar interphalangeal angulation and were all associated with a triphalangeal thumb, while in only one patient a radial deviation associated with a longitudinal epiphyseal bracket of the proximal phalanx was noted.
Three patients (four thumbs) received primary surgical treatment for thumb polydactyly. A three-phalangeal thumb was retained in all. Two of these thumbs were also treated surgically for clinodactyly. Removal of the accessory delta phalanx and reconstruction of the soft tissues was performed in a 3-year-old girl. Follow-up at 18 years of age revealed reduced size of the thumb, limitation of flexion and a secondary radial interphalangeal deviation. A closing wedge osteotomy of the shaft of the proximal phalanx, leaving the delta phalanx undisturbed, was performed in a 12-year-old girl. Follow-up at 18 years of age revealed no loss of thumb alignment but not improved function.
Discussion: The aim of surgical reconstruction of thumb clinodactyly is to create a painless thumb of adequate mobility, stability, alignment and size. Removal of an angulated middle phalanx in a very young child should be associated with a ligament reconstruction from the soft tissues. In an older child or adult, an osteotomy of the shaft of the proximal phalanx or fusion of the oblique joint and realignment with bone resection may be indicated. The real value of a surgical procedure may be evaluated only when the satisfactory result remains unimpaired beyond skeletal maturity.
Conclusion: Surgical reconstruction of thumb clinodactyly may require specific challenges and techniques to obtain a painless, properly aligned, stable thumb with improved function that will retain the satisfactory result beyond skeletal maturity.

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Keywords: Thumb; congenital; clinodactyly; triphalangeal.

1. INTRODUCTION

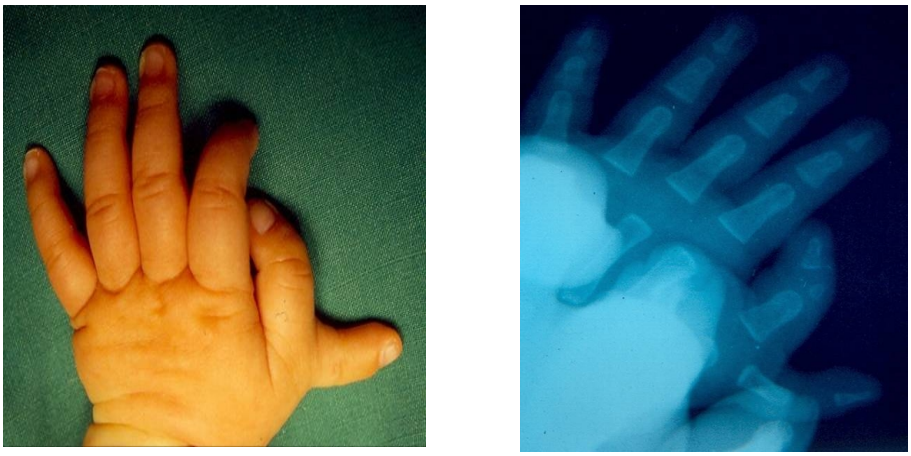
Thumb clinodactyly is a congenital interphalangeal angulation to the radial or ulnar side. The lateral deviation is usually caused by the interposition of an abnormally shaped supernumerary phalanx between the two normal phalanges of the thumb [1] or by a rare ossification anomaly of the proximal or distal phalanx [2,3].

Five patients (six thumbs) with congenital clinodactyly and the results of surgical treatment in two patients, who were followed till skeletal maturity, are presented in this report.

2. CASE PRESENTATION

Five patients (six thumbs) with a congenital thumb clinodactyly have been examined since 1991 (Table 1). There was only one patient with bilateral involvement. In two patients (two thumbs), the anomaly appeared as an isolated lesion. In three patients (four thumbs), it was associated with thumb polydactyly (Fig. 1a). No other dysplasias or generalized syndromes were diagnosed in our patients.

In two of these patients duplication at the metacarpophalangeal joint level was evident and was associated with triphalangism of the ulnar thumb (Fig. 1b); they were classified as type IVB lesions according to the Wassel's classification as suggested by Wood [4,5].



1a **1b**

Fig. 1a and b. A 12-month-old girl with an isolated lesion including thumb polydactyly and ulnar deviation of the ulnar thumb (a) that was due to an extra phalanx (b)

Table 1. Clinical and treatment data

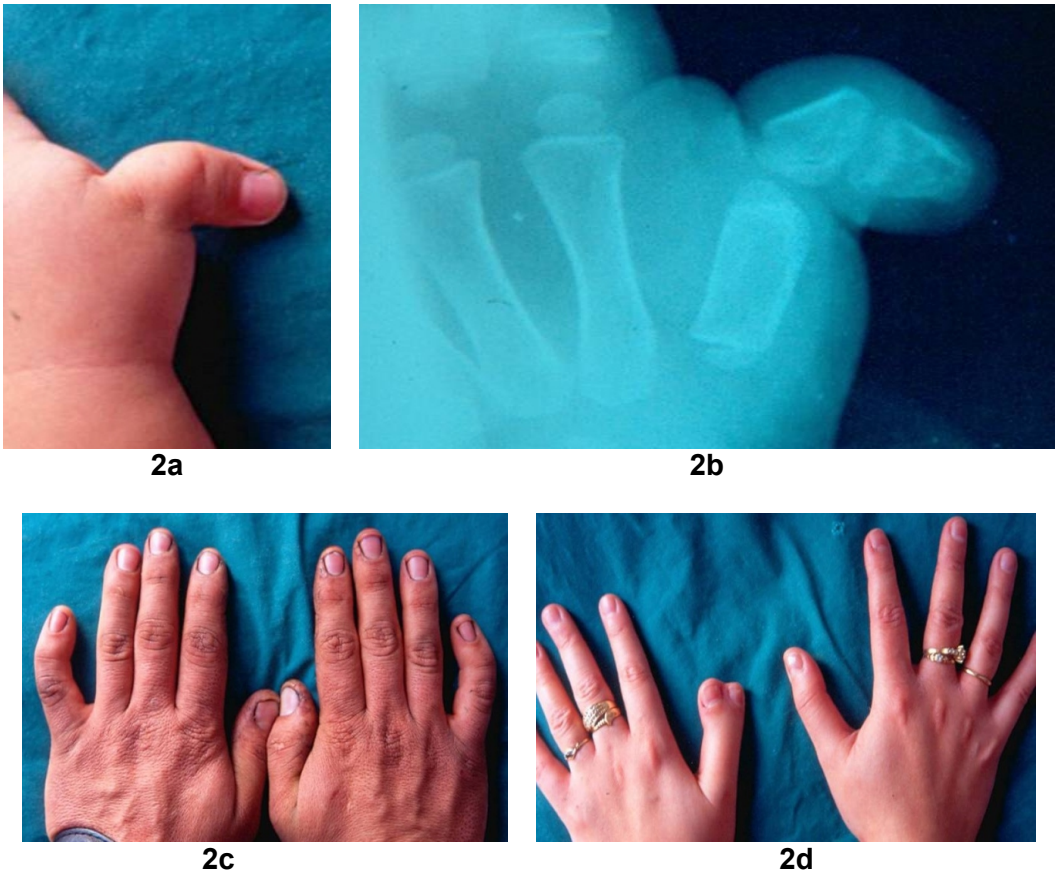
Patient No	Gender	Deviation		Inheritance		Involvement		Surgical Treatment	
		Radial	Ulnar	Familial	Spontaneous	Unilateral	Bilateral	Polydactyly	Clinodactyly
1	F	-	+	-	+	+	-	+	+
2	F	-	+	-	+	-	+	+	+
3	M	-	+	-	+	+	-	-	-
4	F	-	+	-	+	+	-	+	-
5	F	+	-	+	-	+	-	-	-

Key to symbols. + = present; -=absent

Three patients (four thumbs) received surgical treatment for thumb polydactyly at approximately 12 months of age, which included removal of the radial thumb. A three-phalangeal thumb was retained in all. The patient with bilateral involvement was not operated by the author and, therefore, the original type of thumb polydactyly could not be securely established.

In four patients (five thumbs), clinodactyly was associated with a triphalangeal thumb and demonstrated an ulnar deviation. There was only one patient with radial clinodactyly (Fig. 2a) that was due to a longitudinal epiphyseal bracket of the proximal phalanx of the thumb (Fig. 2b).

Four cases were considered to be sporadic, since no family history for similar or other hand abnormalities was noted. There was only one case with diagnosed hand deformities in both parents (Figs. 2c, d).



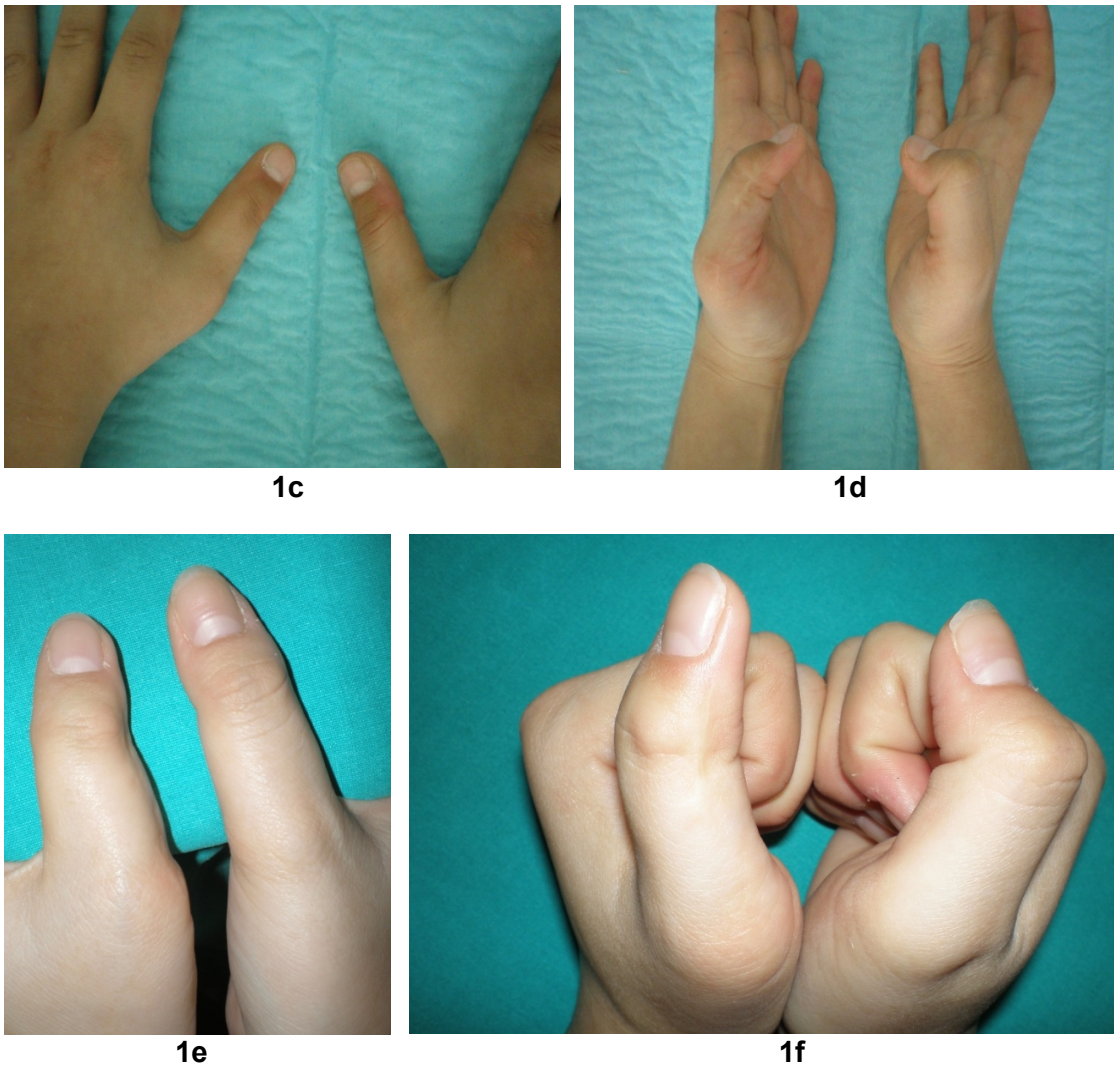
2a **2b**

2c **2d**

Fig. 2(a-d). Radial deviation of the thumb in a 3-year-old girl (a). Radiography indicated a delta proximal phalanx due to a C- shaped epiphysis (b). Her father and mother also presented hand malformations such as bilateral clinodactyly of the fifth finger (c) and duplication of the thumb (d), respectively

Two patients were treated for clinodactyly with ulnarward deviation of the thumb due to a delta phalanx.

In a right-handed dominant 3-year-old girl, the deformity was located on the left thumb. The patient was healthy with no other malformations. Surgical intervention included removal of the triangular accessory phalanx through a lateral incision and collateral ligament reconstruction from the soft tissues and periosteum. The hand was immobilized in a spica cast for 4 weeks. Follow-up at 7 years of age revealed a stable interphalangeal joint with normal anatomical alignment and a 15 degrees reduced arc of flexion (Figs. 1c, d). At 18-years of age a mild radial deviation and a 15 degrees further loss of interphalangeal flexion complicated a painless, stable and functional, although of reduced size, thumb (Figs. 1e, f).

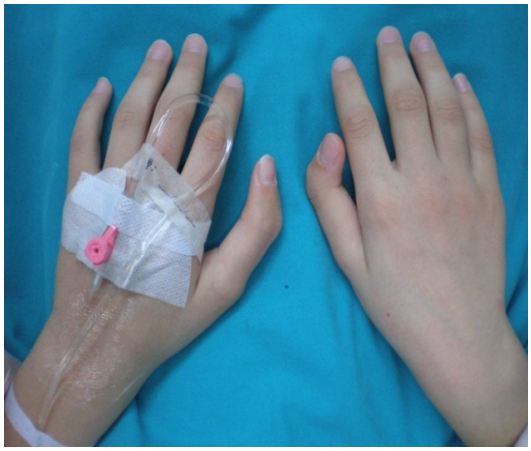


1c **1d**

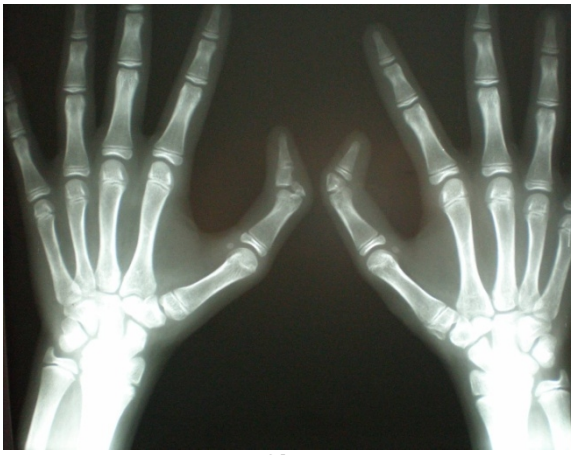
1e **1f**

Fig. 1(c-f). The limitation of flexion of the interphalangeal joint increased between 4 (c, d) and 15 (e, f) years following removal of the delta phalanx

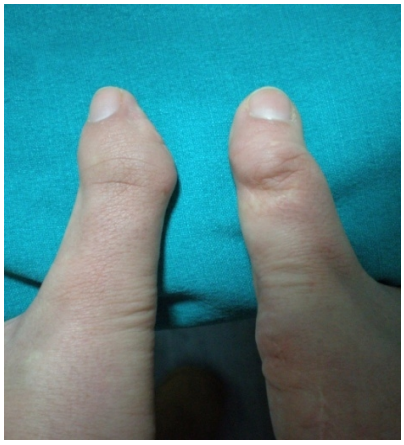
In a right-handed dominant 12-year-old girl, the deformity was located on both thumbs (Figs. 3a, b). Thumb was opposable but strength was diminished bilaterally for all specific thumb functions. She was, otherwise, fit and well. Surgical intervention was performed on the right side and included a closing wedge osteotomy on the convex side of the proximal phalanx fixed with a Kirschner wire. A hand spica cast was used post-operatively for a 5-week-period. Healing of the osteotomy was uneventful. At the last follow-up at 18 years of age alignment of the thumb was not lost but function was not improved. A less pronounced clinodactyly was evident on her left thumb. That was not due to the triphalangeal deformity, since the accessory phalanx was normal (full) in shape and fused at skeletal maturity with the distal phalanx, but to the inclination of the distal phalanx due to the duplication. A significant protuberance, from a piece of the partially removed radial distal phalanx, was evident (Figs. 3c, d). No surgical intervention was undertaken for the deformity on the left side.



3a



3b



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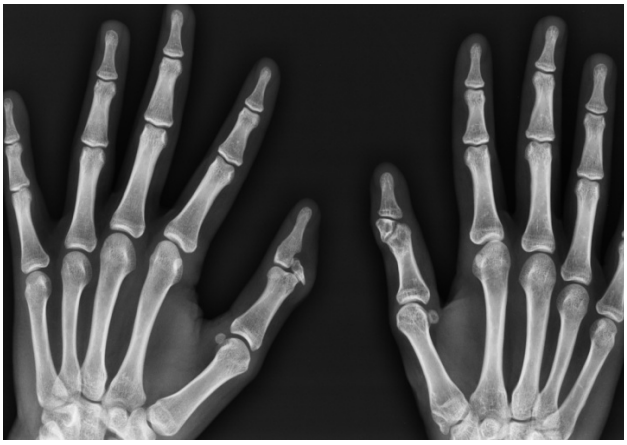

3d

Fig. 3(a-d). Bilateral thumb clinodactyly in a 12-year-old girl. The deformity was more pronounced on the right side (a) that was due to a delta phalanx (b). Clinical and radiographic view 6 years post-operatively (c, d). Bone sclerosis at the distal third of the proximal phalanx of the right thumb indicates the site of the healed osteotomy

3. DISCUSSION

Clinodactyly is a term derived from ancient Greek meaning bent finger and refers to an angulation deformity of a digit in the radioulnar plane [5]. The small finger is the most commonly involved digit, followed by the thumb, the ring finger and most rarely the index and long fingers. The deformity usually involves the distal interphalangeal joint of the small finger bilaterally and exhibits radial deviation [6,7,8].

Classification of clinodactyly may be based on whether the underlying cause is either a soft-tissue or bony abnormality. In the former, angulation may be due to the presence of soft tissue contractures or abnormal fibrous bands on the shorter side as well as to syndactyly [3,9]. In the latter, bony clinodactyly may be due to a triangular appearance usually called “delta” phalanx, named after the triangular Greek letter, or to a trapezoidal deformity of the middle phalanx of a finger caused by a longitudinal bracket epiphysis [10,11].

Clinodactyly may be congenital or acquired secondary to juvenile rheumatoid disease or trauma, frostbite and thermal injury to the growth plate of a digit [3,12]. Congenital clinodactyly caused by both a soft-tissue or bony abnormality may present as an isolated anomaly, usually sporadic or familial, or may be associated with a great variety of genetic syndromes and chromosomal abnormalities [13,14]. An autosomal dominant mode of inheritance with variable expressivity and incomplete penetrance has been confirmed on the majority of familial cases [15,16].

Thumb clinodactyly may be associated with either a two-phalanx or a triphalangeal thumb. In the former, the angular deformity of the thumb may be due to a C-shaped epiphysis or longitudinal epiphyseal bracket of the proximal phalanx, which constitutes a bony bridge that restricts longitudinal growth and causes a triangular deformity of the phalanx and progressive angulation of the thumb [3,17,18]. A triangular-shaped abnormal secondary ossification center of the distal phalanx causing angular deformity of the thumb has also been described [2]. In the latter, thumb deviation is due to an abnormally shaped extra phalanx that may be triangular (delta) or rectangular in shape depending on an early complete or incomplete ossification time of an epiphyseal bracket [5,19].

Triphalangeal thumb may present as an isolated lesion or may be associated with other malformations. The most common one is polydactyly of the thumb. It may also be associated with a variety of syndromes as well as with the thalidomide-caused birth defects. The lesion is inherited in about two-thirds of the patients with triphalangeal thumbs [20,21].

The Wassel’s description of seven types of thumb duplication [4] is the most familiar to pediatric surgeons, since it is a simple classification scheme based on the level of the skeleton at which the bony union-duplication occurs. Any duplication associated with a triphalangeal thumb is described as a type VII lesion. However, it provides no information as to which thumb is dominant and about the presence of soft tissue anomalies [22]. Several classification schemes have been proposed so far in an effort to facilitate communication, analysis and treatment [1,23]

Surgical treatment for thumb polydactyly is usually performed early in life, preferably prior to one year of age and aims to removal of the more hypoplastic and reconstruction of the more developed thumb. When a triphalangeal thumb accompanies a normal or even hypoplastic but functional two-phalangeal thumb, it should be removed. Otherwise, the more functional of the duplicated thumbs is retained.

In this study, three patients (four thumbs) received surgical treatment for thumb polydactyly. A three-phalangeal ulnar thumb was retained in all. In one of them with bilateral involvement exhibiting duplication distal to the proximal phalanx on the left side, surgical removal of the radial distal phalanx excluded a proximal portion probably due to a tendon attachment. Surgery for thumb duplication cannot guarantee an uncomplicated result especially in the more complex deformities, while recombination of the distal phalanx and nail beds in Wassel types I and II malformations has been disappointing [24,25,26].

Surgical excision of the delta phalanx of a triphalangeal thumb should be performed as early as possible and the detached ligament of the interphalangeal joint reconstructed. In an older child or adult, alignment may be achieved by osteotomy of the proximal phalanx or fusion of the oblique joint and realignment with bone resection [1,5,27].

Removal of the accessory delta phalanx and reconstruction of the collateral ligament was performed in a 3-year-old girl. Although a satisfactory result referring to thumb alignment and function was noted four years post-operatively, a long-term follow-up indicated reduced size of the thumb, further limitation of flexion and a secondary radial interphalangeal deviation.

A closing wedge osteotomy of the shaft of the proximal phalanx, leaving the delta phalanx undisturbed, was performed in a 12-year-old girl. A long-term follow-up revealed a painless, stable thumb with no loss of alignment, but no improvement of the movement and strength of the thumb.

Three patients (three thumbs) with clinodactyly did not seek surgical enhancement. Two of them were associated with triphalangism. In the past, surgical treatment for the triphalangeal thumb was not generally advised, since functionality is usually perceived as good. However, it has been recently shown that, although thumb strength is diminished for all specific thumb functions, it is sufficient in daily life. Operative procedures that enhance intrinsic musculature should, therefore, be considered in all types of triphalangeal thumb [28,29]. The ideal time of surgery is suggested to be between 6 months to 2 years [5] or before 6 years of age [30].

4. CONCLUSION

Surgical reconstruction of thumb clinodactyly associated or not with triphalangism may require specific challenges and techniques to obtain a painless, properly aligned and stable thumb with improved function that will not be complicated with secondary deformities or impaired function after skeletal maturity.

CONSENT

Written informed consent was obtained from the patient (or other approved parties) for publication of this case report and accompanying images.

ETHICAL APPROVAL

Not applicable.

COMPETING INTERESTS

The author certifies that he has no commercial associations (such as consultancies, stock ownership, equity interest, patent/licensing arrangements, etc.) that might pose a conflict of interest in connection with the submitted article. The author received no financial support for this study.

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