

## Research Article

### Correction of Congenital Clapsed Thumb Deformity Type 1 & 2: A Case Series

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#### Abstract

**Introduction:** Congenital clapsed thumb is a hereditary flexion and adduction deformity that is akin to a wide variety of structural abnormalities. Timely diagnosis of the deformity is critical for initiating treatment protocol and improving functionality of the hand.

**Objective:** To discuss the treatment protocol of type 1 and type 2 clapsed thumbs and their outcomes.

**Methodology:** This prospective case series was conducted at Department of Plastic & Reconstructive surgery, Dow University of Health Sciences & Dr. Ruth K.M. Pfau Civil Hospital Karachi from June 2016 to May 2020. A total of 10 patients (16 thumbs) with Type 1 and Type 2 clapsed thumb deformity were included; while 4 anomalous cases were excluded. Thorough family, pre-natal and ante-natal history and clinical examination were done. Involvement of unilateral/bilateral hand and disease severity was noted, and data was recorded on a proforma. Type 1 thumb with early presentation was given conservative treatment with splints. Type 2 thumb with web-space contractures was treated surgically with contracture release, tendon transfer and k-wires. Patients were followed for 12 months post treatment.

**Results:** Out of 16 thumbs treated in the study, 5 (31.3%) were given non-surgical treatment while 11 (68.7%) were given surgical treatment. Non-surgical treatment with splint alone was successful in 4 patients with satisfactory results, 1 thumb required surgical intervention later. All 11 thumbs subjected to surgical intervention showed improvement and satisfactory outcomes. 1 required re-correction later.

**Conclusion:** Despite being a progressive disease, congenital clapsed thumb can be managed successfully if timely diagnosis and treatment is initiated. The function of the hand can be drastically improved. Properly planned treatment gives satisfactory results.

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#### Introduction

Congenital clapsed thumb is an advancing flexion and adduction deformity of thumb that may present as part of a syndrome like arthrogryposis, digitolar-dysmorphism, and Freeman-Sheldon syndrome and is associated with various anomalies.<sup>1-3</sup> It may present as a separate entity as well. It is primarily characterized by absence or severe attenuation of extensor pollicis-

brevis (EPB), extensor pollicis longus (EPL) with the involvement of abductor pollicis longus (APL) leading to a tightly flexed, adducted thumb manifesting as varying degrees of first web space contractures and MP joint instability.<sup>4,5</sup>

The diagnosis of this disease can be delayed due to normal position of the thumb inside the palm during first three months of life. However, early diagnosis

and treatment are imperative. If not corrected before cortical functional development, child adapts to the disuse of thumb that severely affects the functionality of hand and the quality of life in the long run.<sup>6,7</sup>

Weckesser et al in 1968 described a classification system of this anomaly into four types.<sup>2</sup> Multiple treatment options are available depending upon the age at presentation, severity of the disease and associated structural deformities. Conservative treatment with splinting provides good functional outcome in supple thumbs that present early, before the age of one year; while surgical correction is reserved for neglected or severe cases presenting with associated web contractures.<sup>8</sup>

The Objective of this study is to discuss the treatment protocol of type 1 and type 2 clasped thumbs and their outcomes.

## Methods

A prospective study was conducted at the Department of Plastic & Reconstructive Surgery Dow University of Health Sciences & Dr. Ruth K. M. Pfau Civil Hospital Karachi, from June 2016 to May 2020. Children aged between 3 months to 3 years with Type 1 and Type 2 clasped thumbs with unilateral or bilateral involvement were included in the study while children with Type 3 and type 4, complicated with heterogenous anomalies in addition to thumb, already defective cortical function (cerebral palsy), severe medical diseases and other systemic or musculo-skeletal congenital anomalies were excluded.

After taking written and informed consent from parents, patients fulfilling the inclusion criteria were enrolled in the study. They were subjected to thorough physical and radiologic evaluation using a proforma by the senior surgeons. Detailed family, prenatal and antenatal history was obtained. All patients were followed for 12 months post-intervention. For the ease of data collection and treatment implementation, deformed thumbs were divided into two main groups based on age at presentation and disease severity (according to Weckesser classification Type 1; supple, and Type 2; complex).

**Group A:** early presentation (before 1 year of age) and supple type 1 deformity

**Group B:** late presentation (after 1 year of age) and complex type 2 deformity.

**Statistical Analysis:** Statistical Package for Social Sciences (SPSS) version 24 was used for the analysis of obtained data.

## Treatment Protocol

**Non-surgical:** Patients who presented early with supple type 1 deformity were offered full time splinting of thumb in extension and abduction for six months. Plaster cast was changed at 6-8 weeks, taking into consideration the growth of the hand. Plaster splint at night was continued for another 6 months after gaining full active extension (Figure-1)

**Surgical treatment:** Surgical intervention was offered to cases that presented late, or with complex type 2 thumbs with joint and web space contractures. Surgery was also offered to cases that failed to respond to splintage alone. The basics of reconstruction included release of the narrowed web space using simple Z-plasty in almost all cases, with 3 cases requiring coverage of the skin defect with a dorsal rotation advancement flap. All the tight structures including the fascia overlying thenar muscles were identified and released. Tendon transfer was done to achieve active extension of thumb at the first Metacarpophalangeal joint. Extensor indicis-proprius (EIP) was the donor tendon used. It was found through a small incision over the dorsum of second MCPJ, tenotomized, re-routed and passed through a subcutaneous tunnel to be transferred to extensor mechanism of thumb or attached to the base of proximal phalanx. If EIP was absent, a slip of flexor digitorum-superficialis (FDS) was used. The thumb was held in extension and abduction stabilizing the MP joint with two cross k-wires.

The hands were kept in a plaster cast and k-wires were removed after 6 weeks. After that, night splint was continued for six months post operatively in extension and dynamic use of thumb was advocated during the day (Figure-2).

## Outcome Assessment

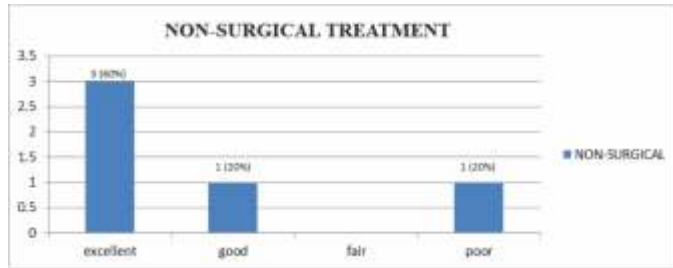
Outcome was assessed on following criteria

- (i) parents' satisfaction regarding appearance and position of thumb
- (ii) opening of the webspace
- (iii) scars
- (iv) function of thumb according to Gilbert's grading system – degree of opposition of thumb during grasping different objects.

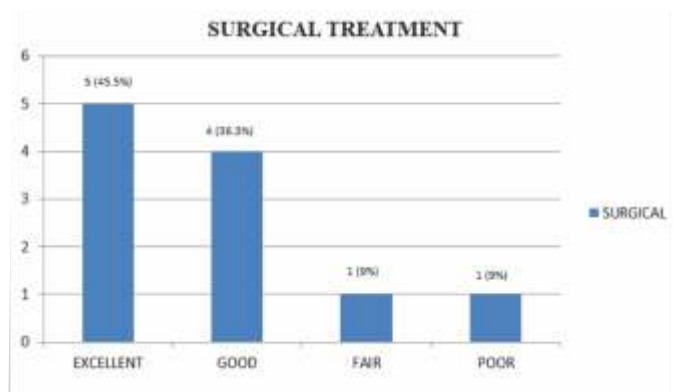
## Results

A total of 10 patients (16 thumbs) were included in the study, seven (70%) patients were male and three (30%)

were female. six (60%) had bilateral thumb involvement and four (40%) had unilateral involvement. Mean age ( $\pm$  SD) was  $14.6 \pm 9.61$  months (range:  $\geq 3$  months – 36 months). All patients were followed for 12 months post treatment. Out of 16 thumbs included in the study, 5(31.3%) were offered non- surgical treatment with splintage while 11(68.7%) were offered surgical intervention. Patient details regarding diagnosis and treatment are elaborated in Table 1.



**Graph 1:** Non-Surgical treatment of Clasped Thumb Deformity



**Graph 2:** Surgical Treatment of Clasped Thumb Deformity

All of our patients’ parents were satisfied with the results as the position of thumb out of the palm alone improved the appearance and functionality of the hand. Functionality was assessed using the Gilbert’s grading system considering the degrees of abduction, rotation and opposition of thumb. Out of 3 patients (5 thumbs) non-surgical treatment with splintage alone was used with excellent outcome in 3(60%) and good in 1 (20%) thumb, such that they could pick up a pencil and key or hold a ball with full active extension of thumb at all joints. Unsatisfactory outcome (poor, according to Gilbert’s chart) was found in 1 thumb (20%), which required surgical correction at alater stage. All 11 thumbs subjected to surgical intervention showed improvement. 5 (45.5%) operated thumbs had excellent, 4(36.3%) had good, 1(9%) had fair and 1 (9%) had poor outcome. Graphs 1 and 2 depict outcomes of non-surgical and surgical intervention for clasped thumb, respectively.



**Figure 1:** Plaster Cast Applied for a Type 1 Deformity with thumb Held in Abduction and

**Table 1:** Patients' thumb involvement, types of deformity and the treatment offered

Thumb involvement	Type of deformity	Treatment
Unilateral	1	Splintage
Unilateral	2	Webspace release(z-plasty), intermetacarpal & adductor fascia release, tendon transfer, k-wiring
Unilateral	2	Webspace release(dorsal rotation advancement flap), intermetacarpal & adductor fascia release, tendon transfer, k-wiring
Unilateral	2	Webspace release(z-plasty), intermetacarpal & adductor fascia release, tendon transfer, k-wiring
Bilateral	1	Splintage
Bilateral	1	Splintage
Bilateral	2	Webspace release(z-plasty), intermetacarpal& adductor fascia release, tendon transfer, k-wiring
Bilateral	2	Webspace release(z -plasty), intermetacarpal fascia release,adductor muscle release from 3 <sup>rd</sup> metacarpal, tendon transfer, k-wiring
Bilateral	2	Webspace release(dorsal rotation advancement flap), intermetacarpal & adductor fascia release, tendon transfer, k-wiring
Bilateral	2	Webspace release(dorsal rotation advancement flap), intermetacarpal fascia release, adductor muscle release from 3 <sup>rd</sup> metacarpal, tendon transfer, k-wiring



### Extension

**Figure 2:** Surgical release of Type 2 deformity. (a) The contracted 1st webspace was released using simple z-plasty (b) EIP tendon was identified and release from its attachment into extension expansion of index finger, re-directed for transfer to the thumb, (c) EIP attached to the base of proximal phalynx and k-wires used to hold thumb in extension and abduction.

### Discussion

Congenital clasped thumb is a hereditary flexion and adduction deformity that manifests as a wide variety of structural abnormalities. First case was described by Tamplin in the early 1840's. It is defined by the lack or attenuation of the extensor mechanism of the thumb with varying degrees of web space contracture and MP joint instability.<sup>1,3,5</sup>

The exact cause of the disease remains unknown but studies have suggested a strong genetic predisposition with incidence of positive family history of 32.5% as reported by Tsuyuguchi et al and Ghani et al®. The disease is more common in males than in females with a ratio of 2.5: 1% as reported by Ghani et al and Lin et al®. with a high incidence of bilateral thumb involvement.<sup>3,9</sup> Similar pattern of inheritance with positive family history and consanguinity was found in about one-third of cases in our study. Male pre-dominance (70%), and high percentage of bilateral thumb involvement (60%) seen in our study was similar to that reported by Tsuyuguchi et al.<sup>10</sup>

Various classification systems have been devised to categorize the disease with its broad spectrum of anomalies. Most widely used classification systems were proposed by McCarroll and Tsuyuguchi. Weckesser et al provided a classification system that divided the disease in 4 types depending on the severity of defor-

mity.<sup>2,8</sup>

We used the classification system proposed by Weckesser et al and our treatment protocol was according to Weckesser et al, Lipskeir and Weizenbluth and McCarroll who divide the deformity in two type (supple –type 1 and complex–type 2 with webspace contractures). Supple type was primarily treated with splintage for up to 6 months with thumb held in abduction and extension. The complex type was offered surgical treatment with correction of the contracture and reconstruction of the extensor mechanism of thumb.<sup>9,11,12</sup>

In this study 31% (5 out 16 thumbs) patients were treated with splintage alone and yielded satisfactory results similar to those reported by Ghani et al and Tsuyuguchi et al, while 68.7% (11 out 16 thumbs) were treated surgically.<sup>8,13</sup> We also noted that the severity of the structural pathologies was mostly dependent on the age of patient at the time of surgery. The corrective and reconstructive options ranged from simple z-plasty in most cases and coverage of the skin defect with dorsal rotation advancement flap to release of adductor fascia and muscle when required. The thumb was held in abduction and extension with the help of two cross k-wires.<sup>14,15,16</sup>

There is no unanimously accepted method for the analysis of treatment outcomes due to difficulty in assessment of function of thumb at the given age.<sup>16</sup> Multiple bench marks have been identified and used in the literature for assessment of success of non-surgical/surgical treatment in terms of function and satisfaction. Some authors assessed degree of active extension of thumb, while Tsuyuguchi et al considered the degree of active abduction at the trapeziometacarpal joint.<sup>11,19,20</sup> Opening of the 1<sup>st</sup> webspace was used as a reference point by Lipskeir and Weizenbluth. We used Gilbert's chart as our benchmark of assessment which is based on active abduction and opposition of thumb.<sup>10,17,21,22</sup> Three out of five thumbs treated conservatively had excellent, one had good, and one thumb had poor outcome as per the grading system used in the study. All 11 thumbs that were treated surgically had satisfactory outcome.

### Conclusion

In this study we found that despite being a progressive disease, congenital clasped thumb can be successfully managed if timely diagnosis and treatment is initiated. The function of the hand can be drastically improved.

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