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Treatment of Common Congenital Hand Conditions

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Abstract

Objective—After reading this article, the participant should be able to: 1. Recognize the clinical features associated with five common congenital hand conditions. 2. Describe the indications and appropriate timing for various surgical procedures used to treat congenital hand anomalies. 3. Identify the pearls and pitfalls of these surgical treatments in order to avoid complications. 4. Understand the expected post-operative outcomes associated with these surgical procedures.

Summary—This article will provide an introduction to congenital hand differences by focusing on practical surgical strategies for treating five commonly encountered conditions including syndactyly, constriction ring syndrome, duplicated thumb, hypoplastic thumb and trigger thumb. The accompanying videos will demonstrate common and reliable surgical techniques for syndactyly release, duplicated thumb reconstruction and pollicization for hypoplastic thumb.

Keywords

Syndactyly; constriction ring syndrome; duplicated thumb; hypoplastic thumb; trigger thumb

Syndactyly

Preoperative assessment

Syndactyly is a common congenital hand anomaly, occurring in approximately one in every 2000 to 3000 live births.(1) Syndactyly can be inherited in an autosomal dominant manner with variable expression or reduced penetrance(2); however, this condition can also occur sporadically. Syndactyly is considered to be complete when the fingers are fused all the way to the tip including the nail folds (Figure 1A); incomplete when the nail folds are not involved (Figure 1B); simple when the fingers are fused by a skin bridge; and complex when the bones are fused together (Figure 1C, D). Syndactyly between the middle and ring fingers is most common, occurring in 57% of the cases, followed by the ring and little fingers,

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which occurs in 27% of the cases.(3) Radiographs of the hand should be taken to confirm skeletal deformities or a concealed extra digit. Syndactyly is often associated with other conditions such as Apert's or Poland syndrome. In these syndromic conditions, the syndactyly is frequently classified as complicated, which is marked by interposing bony structures between the fingers and makes separation more difficult. In these cases, the thumb/index and index/middle digits are often involved.

Goal and indication of surgery

The treatment goal for syndactyly is to create a more normal webspace to improve the function and appearance of each finger.(4) Surgery is generally indicated for all cases of finger syndactyly because the outcome is often better with finger separation. The timing of syndactyly release varies with surgeons, but we will generally perform the separation between 12 and 18 months of age (5). The timing of surgery for simple syndactyly is usually at 12 months or older. Release at an earlier age has a higher incidence of scar contracture, whereas operating later may result in finger deformities as unequal growth of the fingers can cause deviation of the joints. (5-7) The exception to this rule is syndactyly between the ring and the little finger, which requires separation around 6 months of age because the unequal lengths of the fingers can cause finger deviation that can lead to permanent deformity. Conversely, syndactyly between the middle and ring fingers does not require earlier separation because the digits have similar lengths. In Apert's hands, in which there is complicated syndactyly involving all the fingers in the most severe cases, the border digits (thumb/index and ring/little) are separated first to give the child the ability for tripod pinch. In these conditions, we prefer to release the border digits with unequal length around 6 months of age to avoid progressive deviations of the fingers. Only one side of an affected digit should be operated on at one time in order to avoid vascular compromise of the flaps and digits. For syndactyly involving three fingers, the fingers are separated in two different procedures to preserve the vascular supply.

Surgical procedure and complication

For syndactyly correction, it is crucial to understand the anatomy of the webspace. The natural webspace is U-shaped and has a 45-degree slope extending from the metacarpal head to the mid-portion of the proximal phalanx.(4) The index and long webspace is located at approximately the same level. The forth webspace is proximal to the second and third webspace. When viewed from the volar side, the distal extent of the webspace is located at the midpoint between the proximal interphalangeal crease and the distal palmar crease.(1)

The most common procedure for reconstructing the webspace is a proximally-based dorsal rectangular skin flap (Video 1). For complete syndactyly in which the nails are united, laterally based skin flaps from the distal pulp are elevated to create the nail fold (Figure 2). Generally, a full-thickness skin graft harvested from the groin is used to cover the open areas between the separated fingers. A split-thickness skin graft is not preferred because thinner skin grafts have a propensity to contract.

Additionally, some surgeons may attempt to use a dorsal metacarpal flap for syndactyly release. This procedure creates the webspace by using the skin from the dorsal hand and

enables the use of extra finger skin to cover lateral defects on the fingers.(8-10) The major disadvantage of this procedure is the conspicuous longitudinal scar on the dorsum of the hand. In some cases of simple syndactyly, extensive defatting of interdigital space can also allow for closure without skin grafting; however, it is not preferred because defatting can cause nerve injury, venous drainage problems and an emaciated appearance of the finger. (11, 12) Although these procedures may reduce operating time and donor site morbidity, these risks must be carefully considered.

Potential postoperative complications of syndactyly release are related to skin necrosis, skin graft failure and scar contracture, which often require debridement and resurfacing with skin graft.(1) Web-creeping problems can occur as the child grows, so in order to prevent this it is important to release the border digits early and to over-compensate the webspace deepening by recessing the webspace more proximally. In complex syndactyly, joint instability or skeletal deformities can occur. Arthrodesis may be considered to solve the joint instability problem once the child has reached skeletal maturity.(1) Keloid formation following syndactyly release occurs in 1.2% of cases.(13)

Expected outcome

The separation of simple syndactyly successfully improves appearance and preserves mobility. Complex syndactyly release, however, is often associated with loss of mobility due to the higher risk of contracture associated with inherent skeletal deformity.(7)

Constriction ring syndrome

Preoperative assessment

Constriction ring syndrome is a rare condition; the reported incidence ranges from 1/1200 to 1/15000 births.(14-17) The index, middle and ring fingers are frequently affected, whereas the thumb is occasionally involved.(17) Deformities usually occur in multiple extremities and are most predominant in the distal parts.

Constriction ring syndrome is a condition in which the limbs or digits of the fetus become entangled with strands of the embryonic membrane.(17-20) Common features of this syndrome include amputation, acrosyndactyly and lymphedema. The part of the finger distal to the constriction ring is often hypoplastic or absent, whereas the proximal part is intact. Mild constriction ring is often asymptomatic; moderate ring causes lymphedema distal to the ring; and severe constriction ring blocks circulation of the arterial and venous system and causes nerve palsy due to nerve compression.(21) It is necessary to distinguish constriction ring syndrome in an isolated extremity from brachysyndactyly and transverse deficit. Unlike constriction ring syndrome, these anomalies affect only one limb and present with hypoplasia in the proximal part.(22)

Goal and indication of surgery

The purpose of surgery is to release the constriction ring and acrosyndactyly so that the child may have an improved appearance and obtain basic functions such as grip and pinch. Acute exacerbation of the distal edema or nerve palsy is an indication for emergent excision for the

ring to relieve the tension.(23, 24) Distal edema can rapidly progress in the first week after birth. In these cases, expeditious release can resolve this condition. Correction of constriction ring can be performed at 3 months of age, if necessary (25). In cases of acrosyndactyly, surgical treatment is performed between 6 and 12 months of age.(25)

Surgical procedure and complications

Surgical correction of constriction ring includes the excision of the constricted ring and the excessive subcutaneous fibrous tissue and fat. This is followed by z-plasty flaps over the lateral side of the fingers to make the scar less conspicuous (Figure 3).(26) Traditional skin flaps can leave a skin indentation at the ring site because of a deficiency of subcutaneous tissue. A modified technique to advance an adipofascial flap can be useful to fill in the defect (Figure 4).(27) In cases of broad constriction ring, a regional flap, such as a cross-finger flap, may be needed for coverage.(25)

The release of acrosyndactyly follows a similar procedure as syndactyly release. The sinus, which is an epithelialized track, is removed and used as a full-thickness skin graft because it is oftentimes too distal to transform into the webspace.(28) It is easiest to separate digits from proximal to distal end because it is difficult to confirm the commissure at the distal site.

Because all structures proximal to the ring are intact, toe-to-hand transfer and metacarpal lengthening may improve hand function and appearance for distal hypoplasia or absent digit. (29-31) Non-vascularized toe phalanx transfer has also been attempted because it is a simple and safe procedure; however, bone resorption and insufficient growth of the transferred phalanx are common problems.(32, 33)

The most severe postoperative complications of constriction ring release are the potential vascular problems. The distal blood flow is supplied via perforators from the digital arteries and the venous drainage is facilitated by the venae comitantes of the digital arteries (Figure 5).(21) A one-stage operation may fail to preserve the venae comitantes and endanger the digital circulation. Therefore, constriction ring corrections of the digits are best performed in two stages, at least 6 to 12 weeks apart. (34) In the wrist or forearm, one-stage excision of constriction ring is acceptable because deep main arterial and venous vessels are protected. (21, 35)

Expected outcome

The resection of constriction ring and the correction of acrosyndactyly can improve both appearance and function; however, in the case of severe deformity with distal finger amputations and acrosyndactyly, the limited functional benefit with reconstruction should be weighed against the risk of scar formation.(36)

Duplicated thumb

Preoperative assessment

The incidence of duplicated thumb is approximately 1/3000 live births.(37) Preaxial polydactyly is most commonly found in Asians (2.2/1000); however, it affects multiple

ethnic groups including Native Americans (0.25/1000), African Americans (0.08/1000) and Caucasians (0.08/1000).(38-41) The majority of duplicated thumb cases are sporadic, unilateral and do not require genetic consultation; however, it is possible that triphalangeal thumb is associated with an autosomal dominant inheritance pattern.(39, 42, 43)

The Wassel classification system is widely used and classifies thumb duplication according to the level of duplication (Table 1).(44) The extent of hypoplasia, joint stability, and alignment of each thumb influence the choice of the procedure. For type VII thumb duplication (triphalangeal thumb), the treatment can be difficult and is determined by the aberrant shape of the extra phalanx, the narrowing of the webspace between the thumb and index finger, and problem with opposition.(45) Radiographs of the hand are routinely obtained to evaluate the level of the skeletal duplication, hypoplasia and angular deformity of the bones, and the configuration of the articular surface. Observing a child at play, examining for skin creases and comparing the size of both duplicated thumbs are also helpful ways to decide which thumb is dominant. In general, the ulnar thumb is kept to preserve the intact ulnar collateral ligament, which is more important for thumb stability during pinch.

Goal and indication of surgery

All duplicated thumbs will need surgical intervention (Table 1). For the majority of duplicated thumbs, the primary goal of reconstruction is an improved appearance; however, the main treatment goal for triphalangeal thumb is to reconstruct a functional thumb with pinch activity. The ideal reconstructed thumb should be aesthetically adequate in size and have a well-shaped nail plate. Reconstruction is generally performed between 1 and 2 years of age, as pinch function is developing.

Surgical procedure and complications

In addition to osteochondral structural problems, abnormal musculotendinous insertions are also problematic in duplicated thumbs. These common problems result from abnormal points of origin and insertion of the thenar muscles and extrinsic tendons that cause mechanical imbalances in the thumbs.(46) The unusual connection between the flexor pollicis longus (FPL) and the extensor pollicis longus (EPL) tendons on the radial side of the thumb, called the pollex abductus, can cause postoperative angular deformity if not released.(47)

Simple removal of one of the duplicated thumbs will lead to poor long-term results.(48, 49) Instead, resection of the hypoplastic thumb with collateral ligament reconstruction is necessary for treatment of all types of duplicated thumb (Video 2).(50) Despite the best efforts, joint instability and angular deformities can occur as the child grows.(51, 52)

The Bilhaut-Cloquet method consists of central part resection of the bone, soft tissue and nail.(53) Optimal size and appearance can be created by combining outside parts of both thumbs; however, long-term evaluation indicate problems with the growth plate, nail fold and central cleft because it is difficult to precisely match these essential components.(42, 48, 49, 54, 55) Most surgeons perform this procedure for hypoplastic thumbs of Wassel Type I or II only. Beak et al. modified the Bilhaut-Cloquet procedure to preserve the

interphalangeal (IP) joint of the thumb using an extra-articular technique for Wassel Type II and III.(56) The IP joint is reconstructed with one thumb to prevent damage to the physis, and the other thumb contributes part of the distal phalanx for stability (Figure 6). They found that this modified procedure minimized motion loss of the IP joint, nail deformity and growth arrest at a mean follow-up time of 52 months.(56)

For cases of triphalangeal thumb with excessive length or progressive angular deviation, excision of the extra phalanx or arthrodesis of the extra IP joint is required.(57, 58) Arthrodesis is preferred when a child has a larger extra phalanx because joint stability is maintained.

Expected outcome

Satisfactory results can be obtained for Wassel Type I, II and IV, whereas Type III, V, VI and VII are more likely to have unsatisfactory results due to the difficulty of correcting the bony alignment and the instability of the joint.(52, 54) A duplicated thumb that is reconstructed by ablation with ligament reconstruction may be smaller than the contralateral thumb and the motion of the IP or MCP joint may be limited.(42, 52)

Hypoplastic thumb

Preoperative assessment

Hypoplastic thumb can be present in isolation or in combination with any radial deficiency.(59, 60) After duplicated thumb, hypoplastic thumb is the second most frequently encountered thumb anomaly.(47) Bilateral thumb involvement occurs in approximately 60% of children with thumb hypoplasia.(59, 60) The Blauth-Buck Gramcko classification is widely used to describe the hypoplastic thumb and is based on webspace narrowing, hypoplasia of musculoskeletal components, joint instability and abnormalities of extrinsic tendons (Table 2).(61) Hypoplastic thumbs are associated with systemic syndromes such as Holt-Oram syndrome, the vertebral, anal, tracheoesophageal and renal (VATER) anomalies or Fanconi's anemia in 18 to 43 % of the patients.(59, 60) The entire affected upper extremity should be examined to find out the extent of the deficiency over the radial side of the limb.

Goal and indication of surgery

The goal of treatment for hypoplastic thumb is to reconstruct the thumb in order to provide the child with mobility and stability for pinch and grip activity. Depending on the severity of the hypoplastic thumb, different surgical options are indicated (Table 2).(61, 62) Reconstructive surgery should be performed as thumb function develops, which is generally between the ages of 1 to 2 years. Ideally, pollicization should be performed before 1 year of age because this procedure should be completed prior to the development of oppositional pinch; in most cases, surgery certainly should not be delayed after 2 years of age.

Surgical procedure and complication

Reconstruction should achieve 4 goals: (1) widen the thumb-index webspace, (2) stabilize the MCP joint, (3) correct the abnormal extrinsic tendons and (4) provide oppositional

pinch.(61) In order to create a deep and gentle-shaped webspace, 4-flap Z-plasty is commonly used. Stabilizing the MCP joint can usually be accomplished by imbricating the ulnar capsule. In cases of severe instability of the MCP joint, epiphyseal arthrodesis may be required.(63) Abnormalities of extrinsic tendons include anomalous attachment or deficit of the FPL or EPL tendon.(61, 64-66) Tendon transfers using the flexor digitorum sublimis (FDS) from the ring finger and extensor indicis proprius (EIP) can be performed.(61) Pollex abductus, if present, is incised because it hampers active motion of the IP joint and causes radial deviation of the MCP joint.(47)

Transfer of the FDS tendon of the ring finger or the abductor digiti minimi (ADM) muscle (Huber's procedure) are common opponensplasty procedures for the hypoplastic thumb.(64, 67) Because the FDS tendon has appropriate length and is powerful, it can be also used for simultaneous reconstruction of the ulnar collateral ligament of the MCP joint.(64, 68) However, hypoplastic thumb is occasionally associated with an absence or anomaly of the FDS tendons.(69)

We prefer the ADM transfer because the ADM is similar length to the abductor pollicis brevis and opponens pollicis and can restore the appearance of the thenar eminence (Figure 7).(67) Transfer of the origin of the ADM to the palmaris longus tendon creates an appearance close to the natural thenar eminence by shifting the muscle to the more radial position.(70) The ADM often has two tendinous portions. These tendinous portions are transferred via a subcutaneous tunnel to the radial capsule and the imbricated ulnar capsule through the extensor hood at the level of the MCP joint. According to an anatomic study, 17% of specimens have a single ADM insertion site, so in these cases the tendinous portion should be sutured to the extensor hood and ulnar capsule to reinforce ulnar stability.(71) Potential complications of ADM transfer include injuries to the ulnar digital nerve of the little finger, the neurovascular bundle to the ADM or the ulnar nerve and artery during the dissection of the ADM muscle.

Index pollicization is the preferred procedure for severely hypoplastic thumbs, indicated by either an unstable CMC joint or a floating or absent thumb. This method transposes the distal part of the index finger to the thumb position.(Video 3).(72) Because the radial neurovascular bundle of the index finger is occasionally absent in children with hypoplastic thumbs, the ulnar digital artery should be carefully dissected and preserved.

For Type III B condition, vascularized toe-to-thumb or joint transfer has been attempted.(73-75) These procedures give the reconstructed hand 5 fingers; however, the results are often mediocre because of hypoplasia of the thumb, limitation of motion and less power of pinch.

Expected outcome

Opponensplasty by ADM transfer for Type II and III A improves pinch power, dexterity and appearance.(70, 76) Pollicization of an index finger has predictable improvement of hand function.(62, 77, 78) Poor outcomes are associated with Type V or radial deficiency because of poor function of the index finger and other associated anomalies.(79, 80)

Trigger thumb

Preoperative assessment

Trigger thumb in children is characterized by flexion at the IP joint and rarely presents with snapping. In most cases, a nodule or thickening of the A1 pulley is palpable. It is controversial whether trigger thumb found in children is a congenital disorder or acquired after birth. A prospective investigation of 1166 neonates showed no trigger thumb at birth, but 2 cases were observed at a 1-year follow-up.(81) Several other studies have also supported the opinion that childhood trigger thumb is an acquired rather than congenital condition.(82-84) However, cases of trigger thumb associated with trisomy 13 (Patau syndrome), fraternal twins, and families with generational occurrence indicate that there may be a heritable component in certain patient populations.(85, 86)

Goal and indication of surgery

The goal of treatment for trigger thumb is to recover full range of motion at the IP joint. The indication and timing of surgical intervention remains a matter of debate because the rate and age for spontaneous resolution are unclear.(82, 87-90) It was recently reported that 45 of 71 trigger thumbs (63%) spontaneously resolved at an average age of 60.1 months (24-128 months).(91)

Ogino proposes surgery at age of 5 years for persistent triggers.(92) Congenital trigger thumbs almost never cause permanent flexion contracture of the thumb and there is no urgency in performing trigger releases at an early age. We perform surgery after the age of 2-years old, if the trigger persists, because of reduced anesthetic risk and ease of the operation on a larger child.

Surgical technique and complications

Surgery is preformed under general anesthesia. The A1 pulley is longitudinally incised with a V incision at the MCP joint crease. In some cases, the annular pulley distal to the A1 pulley may require division.(93) The smooth excursion of the FPL tendon is confirmed by passive motion at the IP joint. Hypertrophy of the tendon can frequently be seen at the site proximal to the A1 pulley, but this finding is inconsequential.

The radial digital nerve passes over the tendon just proximal to the A1 pulley and with hyperextension of the thumb, this digital nerve is close to the skin and the initial skin incision must not be too deep. The digital nerves are identified and retracted away. The A-1 pulley is divided, under direct vision.

Percutaneous release of tendon sheath using an injection needle is a less invasive procedure for trigger thumb release in children.(94, 95) In order to perform this procedure under local anesthesia, it is essential that the patient understands and cooperates with this operation. The major disadvantage of this procedure is that complete release of the tendon sheath cannot be assured and of course, having a child who will cooperate with local anesthesia is a rarity.

Expected outcome

Tendon sheath release for trigger thumb commonly results in excellent motion of the IP joint. Bowstringing and adhesion is usually not observed, even if the tendon sheath needs to be released beyond the A1 pulley.(88)

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

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Figure 1. Syndactyly. (A) Complete and simple syndactyly of the left ring and little fingers. (B) Incomplete and simple syndactyly of the right middle and ring fingers. (C) Complex syndactyly of the left middle and ring fingers. (D) The radiograph of the case (C).

B

Figure 2.

Nail fold reconstruction. (A) Skin flaps are designed on the tip of digits. (B) The skin flaps are elevated before separation of digits. (C) Each skin flap comes from the adjacent digit and is sutured onto the nail. (Reprinted with permission from Kim, S. E., Chung, K. C. Syndactyly release. In K. C. Chung (Ed.), *Operative technique; hand and wrist surgery*, Vol. 2. Philadelphia: Saunders Elsevier, 2008. Pp. 847-858.)

Figure 3. Z-plasty design on the lateral side of the digit. (Reprinted with permission from Chung, K. C., Kim, S. E. Correction of constriction ring. In K. C. Chung (Ed.), *Operative technique; hand and wrist surgery*, Vol. 2. Philadelphia: Saunders Elsevier, 2008. Pp. 837-846.)

Figure 4.

Advancement of adipose flap. (A) Proximal and distal adipose flaps are elevated from paratenon. (B) Both adipose flaps are advanced into the defect of subcutaneous tissue. (Reprinted with permission from Chung, K. C., Kim, S. E. Correction of constriction ring. In K. C. Chung (Ed.), *Operative technique; hand and wrist surgery*, Vol. 2. Philadelphia: Saunders Elsevier, 2008. Pp. 837-846.)

Figure 5.

Blood supply to the finger. Perforators from digital arteries supply the skin of the finger. Venous return is maintained by venae comitantes around the digital arteries. (Reprinted with permission from Chung, K. C., Kim, S. E. Correction of constriction ring. In K. C. Chung (Ed.), *Operative technique; hand and wrist surgery*, Vol. 2. Philadelphia: Saunders Elsevier, 2008. Pp. 837-846.)

Figure 6.

Illustration of the modified Bilhaut-Cloquet procedure. Black portion is excised in order to preserve the IP joint. (A) For Wassel classification Type II. (B) For Wassel classification Type III. (Modified from Baek, G. H., Gong, H. S., Chung, M. S., et al. Modified Bilhaut-Cloquet procedure for Wassel type-II and III polydactyly of the thumb. *J Bone Joint Surg Am* 89: 534-541, 2007.)

Figure 7.

An 18-month old with a hypoplastic left thumb (Blauth-Buck Gramcko classification Type III A). (A) Preoperative appearance of both hands. The left thumb is hypoplastic and the thenar eminence is absent. (B) Intraoperative findings of abductor digiti minimi transfer. The distal insertion of abductor digiti minimi is detached. (C) Postoperative appearance of the left hand. The thumb is positioned in palmar abduction. (D) Pinch function is restored.

Table 1
The classification of duplicated thumb (44)

Type	Anatomical feature		Frequency	Procedure
Type I	Bifid distal phalanx		4%	Ablation with ligament reconstruction Bilhaut-Cloquet procedure
Type II	Duplicate distal phalanx		16%	Ablation with ligament reconstruction Modified Bilhaut-Cloquet procedure

Type	Anatomical feature		Frequency	Procedure
Type III	Bifid proximal phalanx		11%	Ablation with ligament reconstruction Modified Bilhaut-Cloquet procedure
Type IV	Duplicate proximal phalanx		40%	Ablation with ligament reconstruction
Type V	Bifid metacarpal		10%	Ablation with ligament reconstruction

Type	Anatomical feature		Frequency	Procedure
Type VI	Duplicate metacarpal		4%	Ablation with ligament reconstruction
Type VII	Triphalangia		20%	Excision of extra phalanx Arthrodesis of IP joint Pollicization On-top plasty

Table 2
Classification of Thumb Hypoplasia (61)

Type	Finding	Procedure
Type I	<ul style="list-style-type: none"> Minimal shortening and narrowing 	<ul style="list-style-type: none"> No procedure indicated
Type II	<ul style="list-style-type: none"> Thumb-index web space narrowing Hypoplastic intrinsic thenar muscles MCP joint instability 	<ul style="list-style-type: none"> Reconstruction <ul style="list-style-type: none"> - First-web release - Opponensplasty - UCL reconstruction
Type III	<ul style="list-style-type: none"> Type II features, plus extrinsic tendon abnormalities <ul style="list-style-type: none"> A. Hypoplastic metacarpal, Stable CMC joint B. Partial metacarpal aplasia, Unstable CMC joint 	<ul style="list-style-type: none"> A. Reconstruction <ul style="list-style-type: none"> - First-web release - Opponensplasty - UCL reconstruction - Extrinsic tendon correction B. Pollicization
Type IV	<ul style="list-style-type: none"> Floating thumb 	<ul style="list-style-type: none"> Pollicization
Type V	<ul style="list-style-type: none"> Absent thumb 	<ul style="list-style-type: none"> Pollicization