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Carpometacarpal subluxation and dislocation following first metacarpal distraction osteogenesis

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Abstract
A 4-year-old boy (Case 1) with history of amniotic band syndrome and a 50-year-old man with history of traumatic amputation to right thumb (Case 2) both underwent thumb lengthening procedures with metacarpal osteotomy with distraction osteogenesis. Post operatively, both were noted to have CMC joint instability with volar dislocation in Case 1 and dorsal subluxation in Case 2, which were treated with observation and CMC arthroplasty, respectively. Complications after thumb metacarpal lengthening have been well described throughout literature. Here we present the only reports of CMC subluxation or dislocation following first metacarpal distraction osteogenesis. Clinicians should be aware of this rare but associated complication in patients undergoing this procedure.

Keywords: Carpometacarpal subluxation, metacarpal distraction

Introduction
The thumb accounts for approximately 40% of overall hand function (Moy & Paimer 1992) [9]. Loss of the thumb from traumatic amputation or congenital defect results in significant functional impairment especially with pinch and grasp motions (Chung *et al*. 2015) [2]. Thumb reconstruction is necessary to restore essential hand movements. Among the options for thumb reconstruction is distraction osteogenesis of the thumb metacarpal (Heitmann & Levin 2004) [4]. Complications from thumb metacarpal lengthening include angulation of the lengthened bone and flexion contracture of the adjacent joint especially the MCP joint (Chung *et al*. 2015, Matev 2003) [2, 8]. Previous studies have also reported subluxation or dislocation and delayed union or non-union of the MCP joint (Khan 2012, Nara *et al*. 2017) [10, 5]. To date, there have been no reported cases of carpometacarpal (CMC) joint subluxation or dislocation following thumb metacarpal lengthening.
In this report we present two cases, one pediatric and one adult, of patients who underwent thumb metacarpal distraction osteogenesis and subsequently developed CMC subluxation or dislocation.
Statement of Informed Consent: The patients were informed that data concerning the case would be submitted for publication, and the patients agreed.

Case Report
Case 1: A 4-month old boy was referred to our congenital hand clinic with history significant for amniotic band syndrome complicated by complex syndactyly of the left thumb, index, and long fingers. The left thumb was noted to be amputated through the IP joint level with an acrosyndactyly to the radial aspect of the proximal phalanx of the conjoined index and long fingers (Fig. 1). He initially underwent left thumb and index syndactyl release with excision of polydactylous left index finger at 6 months old (Fig. 2). In an effort to create better length for his thumb, he subsequently underwent pollicization of his left index finger at 12 months old (Figure 3). Postoperatively he was placed in a long arm cast with the thumb pinned, which was removed 5 weeks after surgery. He had an area of wound break down along the medial aspect of his thumb and underwent irrigation and debridement at 15 months old.
He was noted to have a persistently tight first web space despite using his rudimentary foreshortened thumb as his normal thumb as he continued to grow. Given the extremely short nature of his thumb, his parents elected to move forward with first web space release and thumb metacarpal osteotomy and distraction osteogenesis with ex-fix application when he was 4 years old.

After compliance with distraction protocol for 61 days, distraction was stopped, and the ex-fix was subsequently removed 4 weeks later. The patient had gained a total of approximately 3 cm in length (Fig. 4). Unfortunately, on subsequently follow up and postoperative imaging, initially noted at the time distraction was stopped, patient was found to have prominence at the thumb CMC joint with volar CMC joint dislocation (Figure 5). Despite this complication his limited motion, he continued to use the thumb and hand for all activities, including sports and video games. At 10 years old, he was noted to have approximately 20 degrees of painless active range of motion in each direction with circumduction with persistent subluxation as noted on x-rays (Fig. 6). Since the patient was using his thumb quite well without pain, decision was made to continue with observation with the discussion that future CMC fusion to position thumb in more functional position could always be considered down the road.

Case 2

A 49-year-old man sustained a traumatic amputation to his right thumb distal phalanx (Fig. 7). He ultimately underwent Moberg advancement flap after initial revision amputation in the ED. He remained with persistent hypersensitivity, and the decision was made to proceed with revision amputation to the proximal aspect of the distal phalanx at 6 months post op. The patient was fitted for prostheses post operatively; however, after long discussions, he wished to move forward with thumb distraction osteogenesis to restore his 2 cm of thumb length lost in order to be able to adequately complete his pre-injury job function. Thus, at 11 months post injury he underwent right thumb metacarpal osteotomy and distraction osteogenesis with ex-fix application. The ex-fix was left in place for 120 days with a total of 2 cm of distraction gained (Fig. 8).

The patient subsequently underwent open reduction internal fixation of thumb regenerate at 3 months following initial thumb metacarpal lengthening procedure. This post-operative course was complicated by MRSA infection, and he underwent irrigation and debridement with later hardware removal at approximately 6 months following plating of regenerate bone. He was noted to initially have some dorsal subluxation of his thumb metacarpal at his first post-operative visit status post hardware removal (Figure 9). He also developed pain at his thumb CMC articulation with dorsal prominence of thumb CMC joint. Given the continued severe pain at the base of his thumb and persistent joint subluxation on follow up, decision was made to move forward with right thumb CMC arthroplasty in the form of trapeziectomy with FCR tendon transfer, interposition and suspension-plasty without implants 7 months after hardware removal. His thumb was noted to have equal length to contralateral uninjured side with opposition to within 1 cm of small finger distal phalanx and completely to ring finger distal phalanx. Post operatively, the patient progressed with therapy, and at his final follow up visit 8 months after right thumb CMC arthroplasty, he was doing quite well and had returned to work full today.

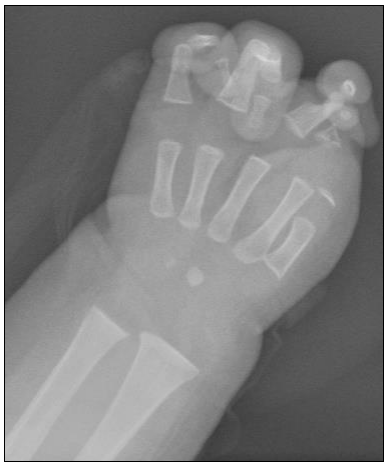

Fig 1: Xrays of the left hand taken at initial visit demonstrating complex complete acrosyndactyly of the thumb, index and long fingers

Fig 2: Postoperative xrays of the left hand after first syndactyly release demonstrating thumb metacarpal with only small, bony remnant of thumb proximal phalanx

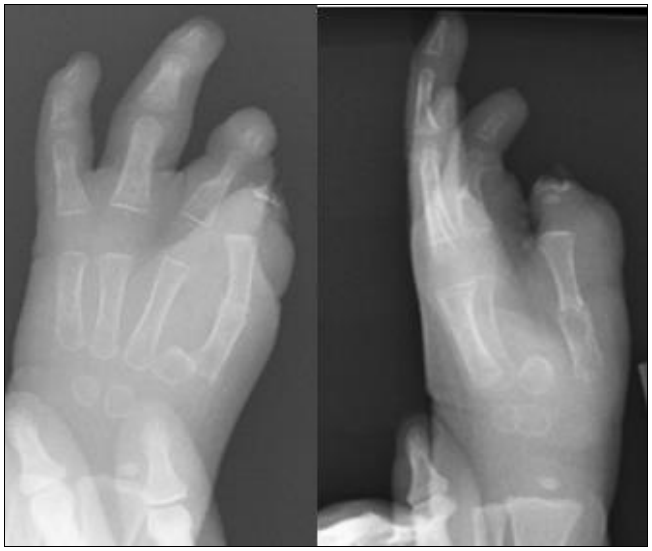

Fig 3: Postoperative xrays of left hand after after index to thumb transposition demonstrating interval healing at thumb index osseous site

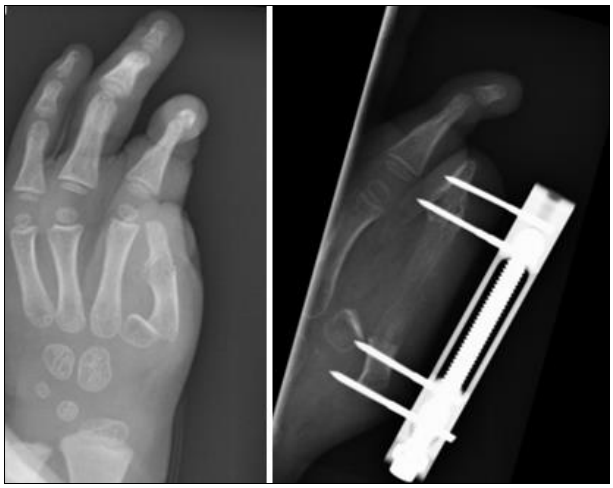

Fig 4: Pre (left) and post (right) thumb lengthening with distraction osteogenesis

Fig 5: Xrays of the left hand at 5 years old demonstrating volarly dislocated thumb CMC joint

Fig 6: Xrays of the left hand at 10 years old redemonstrating persisent thumb CMC joint dislocation.

Fig 7: Initial xrays of right thumb demonstrating thumb amputation through the distal phalangeal level

Fig 8: Pre (left) and post (right) thumb lengthening with distraction osteogenesis demonstrating some regenerate bone at the osteotomy site with approximately 2 centimeters of distraction

Fig 9: Xrays of the right thumb demonstrated healed distraction with dorsal subluxation of the thumb metacarpal on the trapezium

Discussion

In this report we present cases of carpometacarpal subluxation and dislocation following thumb metacarpal lengthening in two patients. Overall, CMC dislocations are an uncommon injury, accounting for 1% of all hand and wrist injuries (Pundkare & Patil 2015) [12]. CMC dislocations of the thumb specifically are rarer compared to CMC dislocations involving the 2nd through 5th metacarpals (Lefere *et al*. 2016) [7]. Furthermore, dorsal dislocation of the thumb is more common than volar dislocation among all CMC joints (Fisher *et al*. 1983) [3]. In the present cases, we describe complications of both volar and dorsal CMC instability. Case 1 presents a case of volar CMC dislocation following thumb lengthening in the setting of congenital deformity, and Case 2 presents a case of dorsal CMC subluxation following thumb lengthening in the setting of traumatic amputation.

The CMC joint of the thumb is made up of the articulation between the trapezium and base of the first metacarpal. The CMC joint of the thumb is surrounded by a capsule and stabilized by ligaments including the anterior oblique, posterior oblique, ulnar collateral, first intermetacarpal, and dorsoradial (Neumann & Bielefeld 2003) [11]. Excessive laxity of any one of these ligaments can lead to instability of the CMC joint and can result in subsequent dislocation. Previous studies have shown that disruption of the

dorsoradial ligament, which connects the dorsal aspect of the trapezium and the first metacarpal, is the most likely cause of CMC joint instability (Lahiji *et al*. 2015) [6]. In Case 1, we speculate CMC volar dislocation was likely due to apex ulnar deviation thumb metacarpal secondary to small healing fracture noted through the regenerate bone on subsequent films after CMC joint dislocation initially observed; whereas CMC dorsal subluxation in Case 2 likely resulted from attenuation of the dorsoradial ligaments as previously described.

Previous studies have shown thumb metacarpal lengthening through distraction osteogenesis to be an effective treatment for thumb reconstruction that is superior to bone grafting (Hietmann & Levin 2004, Toh *et al*. 2002) [4, 13]. This procedure is not without risk (Chung *et al*. 2015, Matev 2012) [2, 8]. Complications include wire tract infection, excessive skin tension, secretion from the openings of the pin bone channels, volar angulation of the lengthened bone, skin graft necrosis, delayed bone union, and flexion contracture of the MCP joint (Matev 2012, Khan 2012, Bari *et al*. 2016) [8, 5, 1]. Flexion contracture of the MCP joint increases the risk for subluxation and dislocation. Multiple studies have reported MCP joint subluxation and dislocation during the metacarpal lengthening of the thumb (Khan 2012, Nara *et al*. 2017) [5, 10]. MCP joint subluxation and dislocation are often seen when mean lengthening percentages were more than 40% of the original length and the lengthening rhythms were greater than 0.5 mm/day (Khan 2012) [5]. Fixation of the MCP joint in extension with a K-wire has been used to prevent flexion contracture and dislocation[4]. Interestingly, MCP joint subluxation or dislocation was not observed in either of our cases. Further analysis shows that in both cases the lengthening protocols were maintained to be lower than 0.5 mm/day and overall length increase was less than 40% likely decreasing the risk for MCP complications.

CMC dislocation is typically managed surgically with primary reduction and thumb Spica casting for 4-6 weeks. Management by anchor suturing and pin fixation with subsequent pin removal at 6 weeks can also be attempted. In Case 1, CMC dislocation was managed conservatively as the patient remained without pain and maintained overall good function of the hand and thumb despite some motion limitation. In Case 2, CMC subluxation was managed with CMC arthroplasty given the persistent pain noted at the base of the thumb, which resulted in inability to return to prior level of work activity.

This case series presents the only report of CMC subluxation or dislocation following thumb metacarpal osteotomy and distraction osteogenesis. CMC dislocation in our pediatric patient was treated conservatively with observation, and CMC subluxation in our adult patient was treated operatively with CMC arthroplasty. While CMC joint instability is a rare complication following thumb lengthening, clinicians should keep a high index of suspicion of this clinical entity in patients undergoing thumb metacarpal distraction osteogenesis.

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