

Case Report

Multiple congenital bilateral trigger digits in a 3-year-old child: a case report and literature review

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Abstract: Trigger finger is a common disorder characterized by clicking, catching, or loss of motion of the involved finger flexor tendon, and is associated with dysfunction and pain. Trigger digits in the pediatric population are relatively uncommon, while multiple congenital bilateral trigger fingers in children are extremely rare. In this paper, we present a 3.5-year-old boy with multiple congenital bilateral trigger digits. Triggering occurred in the proximal interphalangeal joints of the left middle and ring fingers and the right middle finger. The patient was not treated with any physical therapy before his presentation to our clinic. His symptoms completely resolved after surgical treatment with open division of the A1 pulleys. We believe that open release has satisfactory and encouraging results, and may be the preferred method of parents for trigger finger release.

Keywords: Trigger digit, congenital, surgical management, A1 pulley

Introduction

Stenosing tenosynovitis of the digit flexor tendon sheath, also known as trigger finger, is non-infectious inflammation of the flexor tendon sheath, mostly commonly at the A1 pulley, and causes pain, clicking, catching, and loss of motion of the affected finger. Although trigger thumb is a common pathological condition in children, trigger digits are relatively uncommon. Moreover, multiple congenital bilateral trigger fingers in children are extremely rare [1, 2]. In the clinical setting, treatment of trigger finger includes surgical release of the A1 pulley and conservative methods such as steroid injection and splinting. Surgical release is usually recommended when trigger finger persists after conservative treatment [3, 4]. In this study, we report a rare case of bilateral trigger finger in a 3.5-year-old boy that resolved completely with surgical treatment.

Case presentation

The patient was 3.5 years old, and had trigger digits of both hands. His parents stated that triggering was present at birth in his left middle and ring fingers and in his right middle finger

(**Figure 1**). He had no history of perinatal trauma, viral infections, or rheumatologic or metabolic disorders, and had no acute illness or distress. His family members had no history of trigger digits or any other related finger problems. Moreover, he was in good health without any other observable anomaly.

The boy complained of clicking or locking of the affected fingers in the morning, with loosening throughout the day after kneading massage. Physical examination of the affected fingers showed that the proximal interphalangeal joints were locked in a flexed position, and painless nodules could be palpated on the metacarpophalangeal joints of all trigger fingers. Active flexion of these joints was not possible, and passive manipulation was required to achieve full extension. Moreover, passive flexion-extension stretching caused pain and snapping. The sensory examination and peripheral circulation were normal. Radiographs of both hands, and the complete blood count, erythrocyte sedimentation rate, C-reactive protein, and rheumatoid factor levels were all normal.

The operative procedure was performed by one trained orthopedic surgeon. After the patient was given general anesthesia, with the aid of

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Figure 1. The patient had trigger fingers in his left middle and ring fingers and his right middle finger. Triggering occurred at the proximal interphalangeal joints, which were locked in a flexed position, and the patient was unable to perform active flexion of these joints.

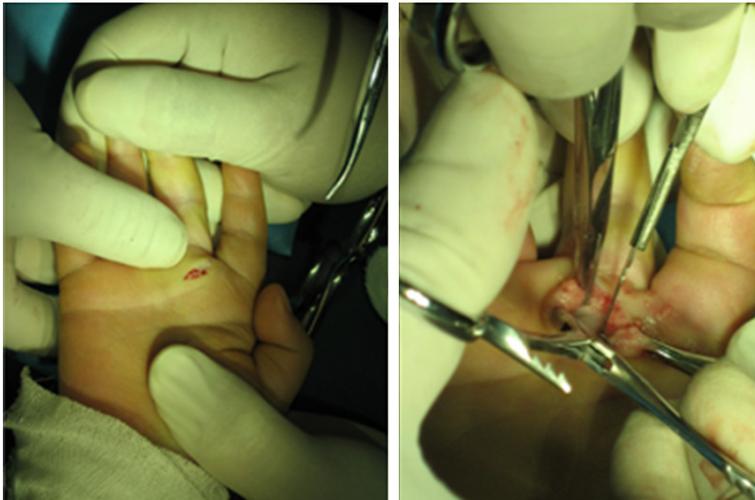


Figure 2. Surgical procedure on both hands - a transverse incision was made on the palm, and releases of the A1 pulleys of the left middle and ring fingers and the right middle finger were performed. A thickened tendon sheath was found, and tendons appeared swollen and roughened, so lateral longitudinal cuts were made perpendicularly to the A1 pulley.

known anatomic landmarks for trigger finger release described by Wilhelmi *et al.* [5], the affected fingers were placed in a fully abducted position. A transverse incision approximately 1-1.5 cm in length was made overlying the proximal edge of the A1 pulley in each affected finger, and blunt dissection was used to fully expose the tendon sheath. We found tendon sheath thickening with no obvious adherence of the superficial and deep layers, and the tendons appeared swollen and slightly roughened on the surface. Lateral longitudinal cuts were made perpendicularly with a scalpel to the A1

pulley (**Figure 2**), until sudden release was felt on the tensioned fingers, the locking was relieved, the snapping disappeared, and the interphalangeal joints could be passively extended. The wounds were then closed in layers. A postoperative dressing was used for 24 hours, and then changed to a bandaid bandage to start passive and active motion earlier. The sutures were removed after 2 weeks. At the 6-month follow-up visit, no signs of infection, nerve injury, finger atrophy, dryness, or reflex sympathetic dystrophy were observed, and the patient was able to flex and extend his fingers without triggering and pain (**Figure 3**).

Discussion

Trigger finger was first described by Notta in 1850, and it is a noninfectious inflammation of the flexor tendon sheath [6]. Trigger digits in infants and children are relatively uncommon. The reported incidence of trigger digit in children varies from 1-6 out of 2000 live births. Boys and girls are equally affected [7]. While trigger thumb is more common, triggering of the other fingers is rare. Moreover, multiple congenital bilateral trigger fingers in children are extremely rare [1, 2]. However,

the precise etiology of trigger finger has not been elucidated, and many factors have been proposed to be associated with this pathological condition [2]. It has been demonstrated that the osseous-fiber tunnel at the metacarpal head area, thickened tendons and tendon sheaths, trauma, metabolic disorders, mucopolysaccharidosis, and genetic factors may contribute to the condition [2].

Controversy remains as to whether trigger digit in children is a congenital or an early acquired condition [8, 9]. Dinham and Meggitt claimed

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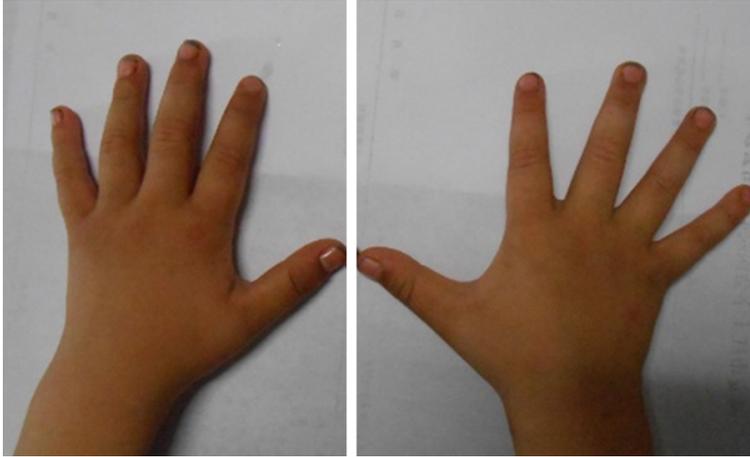


Figure 3. Six months after the operation on both hands. Full extension of the operated fingers was present without triggering or pain.

that trigger digit was present at birth [8], while Moon *et al* examined 7700 newborn children and found no signs of triggering at birth [7], and reported that trigger digit develops during early childhood. Sharma *et al* reported that the mean age at the onset of trigger fingers in children is 11 months [4]. In our case, the patients' parents claimed that he had trigger finger since birth.

In trigger finger, inflammation and hypertrophy of the retinacular sheath restricts both flexion and extension of the flexor tendon. Digit pulleys serve to maximize the flexor tendon's force production and efficiency of motion. Although Akhtar *et al* reported that trigger fingers affected the A2 and A3 pulleys [10], the A1 pulley at the metacarpal head area is subjected to the highest forces and pressure gradients, so it is most often affected in trigger fingers [11]. Pathological examination of the A1 pulleys of trigger digits showed evidence of degeneration and inflammatory cell infiltration, which was characterized by chondrocyte proliferation and type III collagen production coating the pulley's entire innermost layer [12, 13].

The treatment of trigger digits remains controversial. For those cases without snapping or locking, trigger digits may recover spontaneously within 6 months [8], whereas for cases with snapping or locking, the interphalangeal joints of trigger fingers are locked in a flexed position, and this may lead to contracture of the joint capsule and collateral ligaments. Furthermore, for those cases that are not given

effective treatment beyond 6 months, maldevelopment of the bones may occur, so treatment is more urgent. There are several methods of treatment, including conservative treatments such as steroid injection and splinting, with splinting having a reported success rate between 50% and 65% [14, 15]. Pargali *et al* reported that they treated a bilateral trigger finger case with physiotherapy (eight sessions) and non-steroidal anti-inflammatory drugs (for 20 days) [2], and the symptoms completely resolved within 4 months of onset. Unlike in

adults, steroid injection is not suitable for children, as it may cause adherence surrounding the flexor tendon. Operative treatment, whether by percutaneous or open release, is highly effective and has a low complication and recurrence rate, and is widely regarded as the optimal treatment for trigger finger. Both percutaneous and open release methods aim to release the entrapped tendon and fully incise the A1 pulley [16]. However, the timing of surgery remains a topic of debate. Sevencan recommended surgical release for those cases that presented or persisted after 1 year of age [17]. De Luna *et al* recommended surgical treatment after 3 years of age [1]. Open release provides greater exposure and may be safer with regard to iatrogenic neurovascular injury, with success rates from 60% to 100% [18-20]. In our case, the patient's bilateral trigger fingers had persisted for more than 3 years. We cut his affected A1 pulleys using an open release, and locking and snapping were completely resolved after surgical treatment. Six months postoperatively, the outcome was satisfactory, as he could fully extend the operated fingers with no triggering or pain, and had no signs of recurrence.

In conclusion, we recommend treatment for trigger finger at an early age. Considering the potential complications of percutaneous release—such as damage to the neurovascular bundles, injury or inadvertent release of the A2 pulley and flexor tendon, and bowstringing after A2 pulley injury [21]—we believe that open

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release is the preferred treatment method in trigger fingers that have persisted for more than 6 months and show no signs of spontaneous recovery.

Disclosure of conflict of interest

None.

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References

- [1] De Luna V, Potenza V, Garro L, Farsetti P and Caterini R. Multiple congenital bilateral trigger digits in a 2-year-old child: case report. *Open Orthop J* 2013; 7: 75-77.
- [2] Pargali N and Habibzadeh F. Bilateral trigger finger in a 5-year-old child: case report. *J Plast Reconstr Aesthet Surg* 2011; 64: e283-284.
- [3] De Smet L, Steenwerckx A and Van Ransbeeck H. The so-called congenital trigger digit: further experience. *Acta Orthop Belg* 1998; 64: 306-308.
- [4] Sharma PR, Gore SM and Schreuder FB. Bilateral trigger finger in a 7-year-old after a viral infection: case report. *J Hand Surg Am* 2010; 35: 1334-1335.
- [5] Wilhelmi BJ, Snyder Nt, Verbese JE, Ganchi PA and Lee WP. Trigger finger release with hand surface landmark ratios: an anatomic and clinical study. *Plast Reconstr Surg* 2001; 108: 908-915.
- [6] Notta A. Recherches sur une affection particuliere des gaines tendineuses de la main. *Arch Gen Med* 1850; 24: 142.
- [7] Moon WN, Suh SW and Kim IC. Trigger digits in children. *J Hand Surg Br* 2001; 26: 11-12.
- [8] Dinham JM and Meggitt BF. Trigger thumbs in children. A review of the natural history and indications for treatment in 105 patients. *J Bone Joint Surg Br* 1974; 56: 153-155.
- [9] Slakey JB and Hennrikus WL. Acquired thumb flexion contracture in children: congenital trigger thumb. *J Bone Joint Surg Br* 1996; 78: 481-483.
- [10] Akhtar S, Bradley MJ, Quinton DN and Burke FD. Management and referral for trigger finger/thumb. *BMJ* 2005; 331: 30-33.
- [11] Lin GT, Amadio PC, An KN and Cooney WP. Functional anatomy of the human digital flexor pulley system. *J Hand Surg Am* 1989; 14: 949-956.
- [12] Sbernardori MC, Mazzarello V and Tranquilli-Leali P. Scanning electron microscopic findings of the gliding surface of the A1 pulley in trigger fingers and thumbs. *J Hand Surg Eur Vol* 2007; 32: 384-387.
- [13] Sampson SP, Badalamente MA, Hurst LC and Seidman J. Pathobiology of the human A1 pulley in trigger finger. *J Hand Surg Am* 1991; 16: 714-721.
- [14] Rodgers JA, McCarthy JA and Tiedeman JJ. Functional distal interphalangeal joint splinting for trigger finger in laborers: a review and cadaver investigation. *Orthopedics* 1998; 21: 305-309; discussion 309-310.
- [15] Patel MR and Bassini L. Trigger fingers and thumb: when to splint, inject, or operate. *J Hand Surg Am* 1992; 17: 110-113.
- [16] Ryzewicz M and Wolf JM. Trigger digits: principles, management, and complications. *J Hand Surg Am* 2006; 31: 135-146.
- [17] Sevenscan A, Inan U, Kose N, Omeroglu H and Seber S. Percutaneous release for trigger thumbs in children: improvements of the technique and results of 31 thumbs. *J Pediatr Orthop* 2010; 30: 705-709.
- [18] Bonnici AV and Spencer JD. A survey of 'trigger finger' in adults. *J Hand Surg Br* 1988; 13: 202-203.
- [19] Turowski GA, Zdankiewicz PD and Thomson JG. The results of surgical treatment of trigger finger. *J Hand Surg Am* 1997; 22: 145-149.
- [20] Hodgkinson JP, Unwin A, Noble J and Binns MS. Retrospective study of 120 trigger digits treated surgically. *J R Coll Surg Edinb* 1988; 33: 88-90.
- [21] Makkouk AH, Oetgen ME, Swigart CR and Dodds SD. Trigger finger: etiology, evaluation, and treatment. *Curr Rev Musculoskelet Med* 2008; 1: 92-96.