

Thumb Hypoplasia Type IIIA: A Case Report

Phan Duc Minh Man¹, Nguyen Duong Phi^{2#}, Mai Trong Tuong¹, Nguyen Vo Dang Khoa³ and Truong Quang Dinh^{2#}

¹Hospital for Trauma and Orthopaedics, Ho Chi Minh City, Viet Nam

²City Children Hospital, Ho Chi Minh City, Viet Nam

³Thu Duc District Hospital, Ho Chi Minh City, Viet Nam

Abstract

Thumb hypoplasia occurs in a variety of forms, which are often categorized using Blauth classification and its modifications. In 1991, Manske et al. suggested a type IIIA variation characterized by first web space narrowing, underdeveloped or absence of thenar muscles, thumb extrinsic and metacarpophalangeal joint instability. Here we present a 5-year-old female patient who visited our clinic due to a small thumb and inability to flex the interphalangeal joint since birth. Her medical history showed an esophageal atresia surgery which was performed when she was a neonate. She then underwent a first web space release using four-flap Z-plasty and exploration of the flexor pollicis longus tendon. We found that the extensor pollicis longus and flexor pollicis longus tendon were interconnected along the radial border of the thumb, traction on either tendon tends to abduct the thumb at the metacarpophalangeal joint. Tenolysis was performed and the patient showed significant improvements in opposition movements and precision grip post-operation. We proposed that ultrasound is as reliable as magnetic resonance imaging to examine the flexor tendon and performing a wrist incision to confirm the recovery of the thumb opposition motion can save unnecessary tendon transfer.

Keywords: Hypoplasia • Tenolysis • Syndrome

Introduction

Thumb hypoplasia accounts for approximately 5% to 15% of congenital hand disorders [1,2]. The condition occurs equally among both genders. In over half of the cases, both thumbs are affected, while among one-sided hypoplastic patients the right thumb is reported to be more commonly involved [3,4]. Forty percent of cases are associated with known syndromes such as radial longitudinal dysplasia, VACTERL syndrome (vertebral defects, anal atresia, cardiac defects, tracheoesophageal fistula, renal anomalies, and limb abnormalities), Fanconi anemia, Thrombocytopenia-absent radius, Holt-Oram syndrome [3,5]. Thumb hypoplasia is characterized by shrinking in the thumb size, metacarpal adduction, metacarpophalangeal joint instability, thenar muscles hypoplasia and extrinsic tendons dysplasia. Metacarpophalangeal (MCP) joint instability or thumb aplasia are presented in the most severe cases, which are recommended to be treated using thumb removal surgery and pollicization of the index finger. Also, pollex abductus should be aware of when there are no creases noted in the volar plate [6]. Classification systems of thumb hypoplasia assist surgical decision making and reflect the progression in the understanding of this condition. While Müller was among the first to categorize thumb hypoplasia, Blauth's classification (and Buck-Gramcko's modification) is the most used categorization. The principle of hypoplastic thumb reconstruction is to address each abnormal clinical element. The first web space should be deepened [6], the MCP joint stabilized, and the opposition power restored [7]. Generally, little can be done to augment the thumb size and Interphalangeal (IP) joint stiffness, therefore the patients and their parents should be informed that the thumb might continue to be smaller in appearance and the stiff IP joint remains as an unsolved problem, even with successful surgical reconstruction.

***Address for Correspondence:** Truong Quang Dinh, Director of City Children Hospital, City Children Hospital at Ho Chi Minh City, Viet Nam, E-mail: truongdinhnd2@gmail.com

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Received 08 December 2020; **Accepted** 18 January 2021; **Published** 25 January 2021

Case Report

A 5-year-old girl was admitted to our hospital because of a small and less functional right thumb (Figure 1). She could not hold a pen properly and her handwriting was unreadable that had significant negative impacts on her study. This condition was recognized since birth by her mother. Medical history showed a tracheoesophageal fistula surgery in the first month of the patient's life. Physical examination showed no skin creases on the right thumb, which is also limited in size in comparison with her left thumb (Figure 2).

The patient is unable to flex her IP joint actively, although approximately



Figure 1. Comparison between two hands, the right thumb is significantly smaller.

45-degree of flexion can be archived with passive motion (Figure 3). We then carefully examined the thumb MCP ulnar collateral ligament and found no abnormality. The thumb opposition motion is limited (Figure 4). X-ray results showed mild hypoplasia of the carpus bone (Figure 5) and ultrasound confirmed that the flexor tendon still exists.

We decided to widen the first web space using four-flap Z-plasty (Figure 6) and explore the Flexor Pollicis Longus (FPL) using volar incisions. We had no difficulty in widening the first web space during the initial surgery. During volar exploration, we found multiple interconnections between the FPL and the Extensor Pollicis Longus (EPL) and the FPL which was deviated from the ulnar side (Figure 7) causing her disability of flexing the IP thumb actively and weak opposition motion. After the successful tendon separation, we made another incision on her wrist to search for the FPL tendon. We pulled the FPL tendon to confirm that the thumb motion is recovered. Finally, we sewed her subcutaneous tissues and skin using absorbable sutures. Her upper limb was placed in an above-elbow splint postoperatively for 4 weeks. After removing the splint, the patient was referred to a physiotherapist for mobilization exercises in a month. Although the IP joint flexion was not improved, the patient can grasp her hand effectively and showed significant improvement in her handwriting (Figure 8).



Figure 2. No crease on the right thumb.

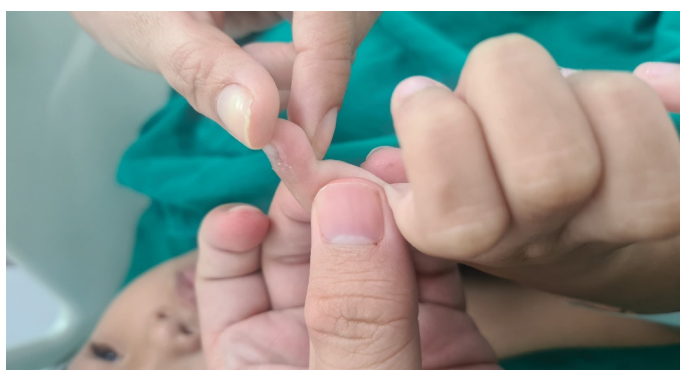


Figure 3. Passive flexion of the right thumb.



Figure 4. The thumb can only meet the index finger when doing opposition motion.



Figure 5. X-ray of the case.



Figure 6. Widening the first web space.

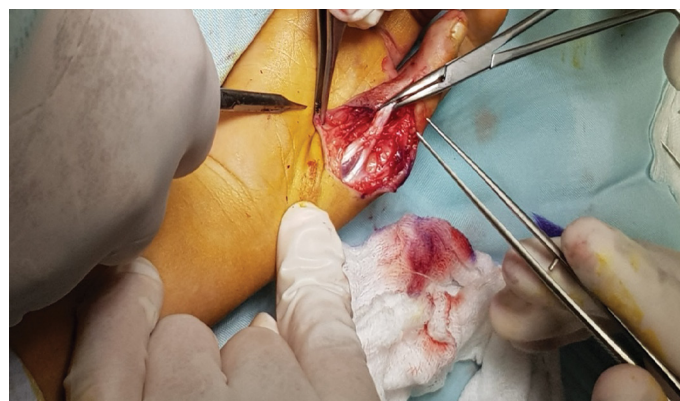


Figure 7. Tendons adhesion between the FPL and the EPL.



Figure 8. Significantly improvements obtained post-operation regarding opposition motion, the thumb can now reach the ring finger.

Discussion

The primary goals of hypoplastic thumb treatment are to stabilize and recover the thumb's opposition motion. The identification and treatment of extrinsic tendon abnormalities are therefore important, especially in thumb hypoplasia type IIIA. Pollex abductus, the interconnection between flexor and extensor tendons, is widely recognized and reported in previous papers [8]. Manske et al. observed the interconnection in 2 of 13 thumbs (15%) [9], while Lister reported in 11 of 31 thumbs (36%) [1], and Graham and Louis observed in 14 of 14 thumbs (100%) [10]. Yuki Bessho et al. illustrated several cases of thumb hypoplasia type IIIA in 16 years from 2001 to 2017, of which 50 out of 79 thumbs had interconnections of FPL and EPL that resulted in an imbalanced force between the flexion and extension of the thumb [11]. Consequently, on account of the FPL radial deviation, the hypoplasia thumb is incapable of not only flexing actively but also limiting the thumb opposition.

In this case, we found that ultrasound can replace magnetic resonance imaging to be utilized as a cheap, simple, non-invasive and effective technique in the evaluation of the tendons to prepare for surgical plan with acceptable accuracy. Such affordable diagnostic technique can be widely applied among developing countries to confirm the existence of the flexor tendon which is a major contributor to decide the surgical approach. However, since ultrasound is often considered as a subjective evaluation that depends on the experience of the sonographer, further research is required to comprehensively investigate the reliability and validity of the technique and propose a systematic approach to ensure diagnostic precision.

After separating the tendons and widening the first web space, surgeons generally perform tendon transfer [12], which usually require a longer time to recover and increase the risk of several potential complications such as infection, damage to adjacent blood vessels or nerves, rupture or loosening of the tendon. Here we propose a new but simple method to examine the recovery of thumb movement after tenolysis through wrist incision. Surgeons can pull the FPL tendon to check the opposition motion. If the thumb opposition is good enough, tendon transfer may not be necessary. Performing such incision can add valuable information to support decision making for physicians.

Conclusion

The priority aim of thumb hypoplasia treatment is to recover the opposition

motion. We proposed that ultrasound is as reliable as magnetic resonance imaging in examining the flexor tendon pre-operation, and performing a wrist incision after tendon separating and first web space widening to confirm the recovery of the thumb opposition motion can save unnecessary tendon transfer for patients.

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How to cite this article: Phan Duc Minh Man, Nguyen Duong Phi, Mai Trong Tuong, Nguyen Vo Dang Khoa, et al. "Thumb Hypoplasia Type IIIA: A Case Report." *Clin Case Rep* 11 (2021): 1412.