

Case Report

Bilateral isolated absence of flexor pollicis longus tendon: report on a rare case management

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ABSTRACT

Introduction: In human, the flexor pollicis longus (FPL) is an important muscle for function of the hand. Thumb aplasia and hypoplasia represent a large spectrum of deficiencies. One of the variants of type I thumb hypoplasia is the absence or hypoplastic of FPL tendon without deficiency of other thenar muscle. Although rare, we found several literatures reporting the cases. In treating pediatric thumb disease especially for this condition, we have to carefully diagnose the problem to set our goal of treatment.

Methods: We presented a case of 8-year-old male with bilateral inability to flex both thumbs at the interphalangeal (IP) joint. His parents realized this condition after he was involved in school activity. On physical examination, we concluded that the patient has absence of FPL and proceeded for reconstruction with tendon transfer from 4th flexor digitorum superficialis (FDS) and dorsal capsule release.

Results: We performed surgery and postoperatively observed the active movement of IP joint. The treatment improved the active flexion of the IP joint around 60 degrees.

Conclusion: The absence of IP crease is one of the clear signs of FPL absence or hypoplasia. The usage of dorsal capsule release and tendon transfer in treating FPL absence or hypoplasia without any other thenar anomalies is proven to be adequate and feasible

ABSTRAK

Pendahuluan: Pada manusia, flexor pollicis longus adalah otot yang sangat penting terhadap fungsi tangan. Thumb aplasia dan hypoplasia memiliki spektrum defisiensi yang sangat luas. Salah satu varian dari thumb hypoplasia tipe I adalah ketiadaan atau hipoplasia dari tendon FPL tanpa defisiensi otot-otot thenar lainnya. Walaupun jarang, kami menemukan beberapa literatur melaporkan kasusnya. Dalam menatalaksana kelainan jempol pada pediatri terutama pada kasus seperti ini kita harus berhati-hati dalam menegakkan diagnosis dan menetapkan target daripada tatalaksana kita.

Metode: Kami mempresentasikan sebuah kasus seorang anak laki-laki berumur 8 tahun dengan ketidakmampuan dalam menekuk kedua jempol pada sendi interphalangeal (IP). Orangtuanya menyadari kondisi ini setelah pasien terlibat dalam kegiatan sekolah. Pada pemeriksaan fisik kami simpulkan pasien mengalami ketiadaan FPL dan kami berencana untuk dilakukan rekonstruksi dengan tendon transfer dari flexor digitorum superficialis (FDS) ke 4 dan dorsal capsule release.

Hasil: Setelah kami lakukan operasi kami dapatkan adanya gerakan aktif dari sendi IP. Tatalaksana dari kami meningkatkan fleksi aktif dari IP hingga 60 derajat.

Kesimpulan: Ketidadaan dari garis tekukan kulit pada IP merupakan salah satu tanda jelas daripada ketiadaan atau hipoplasia daripada FPL. Penerapan dorsal capsule release dan tendon transfer dalam menatalaksana ketiadaan atau hipoplasia daripada FPL tanpa adanya keterlibatan atau kelainan otot-otot thenar lainnya terbukti cukup adekuat dan mampu laksana.

Keywords: congenital isolated absence, flexor pollicis longus, tendon transfer

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INTRODUCTION

In man, FPL is an important muscle for the function of the hand. The thumb is the most important part of the hand because of its strength, which is equal to that of all fingers. Approximately 50% of the function of the hand is attributed to the actions of the thumb. Froment in 1895 was the first to report agenesis of the FPL, which was associated with lumbrical anomalies, absent thenar muscles and absent FDS. Isolated aplasia or hypoplasia of FPL is an anatomical rarity. One of the variants of type I thumb hypoplasia is the absence of FPL tendon without deficiency of other thenar muscle. Although rare, we have several literatures reporting the cases and only two cases documented with bilateral abnormality. Many of the patients rejected surgical intervention.¹⁻³

METHODS

An 8 year-old boy was brought to our clinic for examination of both his hand. He was unable to flex both of his thumb at the IP joint but his parents only realize recently when he was more involved in school activity. He could perform tasks like writing or holding objects. There were no history of trauma and family with similar abnormality. On physical examination, both thumbs are the same size with the absence of IP crease (Fig1).

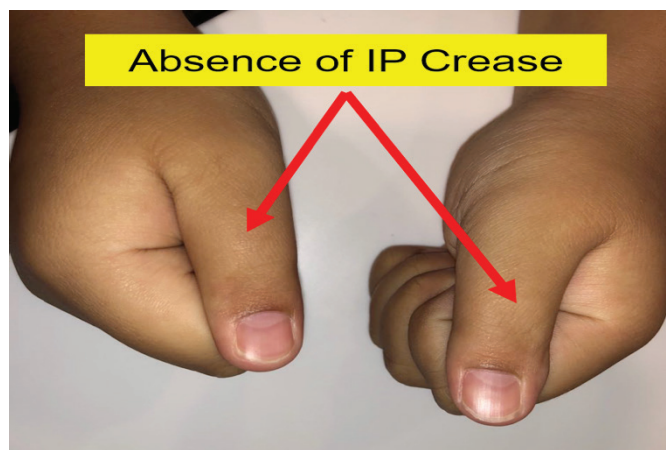


Figure 1. The thumbs with the absence of interphalangeal crease

The patient has a history of surgery for his right thumb. At the time, the attending physician diagnosed the patient with congenital trigger thumb. The patient was then referred to our hand clinic for the treatment of the left thumb. Currently, the right thumb has IP joint stiffness and passive flexion of 10 degree.

Treatment options are one or two stages of tendon transfer and fusion of IP joint, in functional position, in case of weakness or painful movements of the thumb. There was no significant difference in outcome between the one or the two stages of tendon transfer, so we chose to do the one stage surgery. We decided to perform dorsal capsule release and tendon transfer from the 4th FDS. The FDS IV tendon is cut distal to the chiasma of Camper, pulled through the carpal tunnel and moved into the channel of the FPL tendon. Intraoperatively, we found a thin tendon at the distal end with no connection to the proximal band and then wove it using the Pulvertaft technique with the FDS IV tendon at the base of the distal phalanx of the thumb (Fig 2).^{4,5}

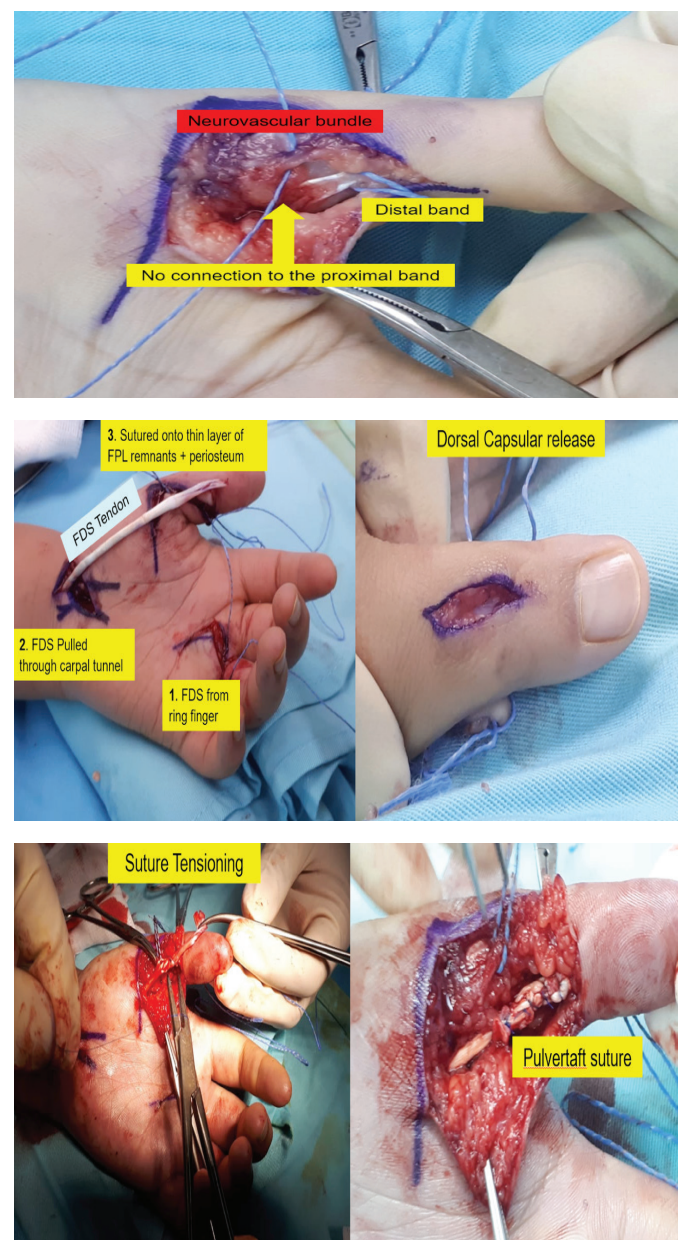


Figure 2. The steps in the surgery

RESULTS

We succeeded performing the surgery, and postoperatively, we observed the active movement of the left thumb IP joint, which was then increased up to 60 degrees compared to the contralateral with no active flexion and 10 degrees passive flexion. At 2 months follow-up there was no complaint from the patient and the patient was able to pinch using the left thumb with the flexion of IP joint.

DISCUSSION

Congenital inability to flex the IP joint of the thumb may be due to several causes, including congenital absence of the FPL, anomalous insertion of FPL, congenital tenovaginitis of the flexor tendon sheath, partial anterior interosseous nerve paralysis, traumatic rupture of FPL, and anomalous band connecting tendons. A literature by Dheer, *et al.*, also illustrated the case of hypoplastic FPL which was firstly thought as FPL tendon rupture, the patient was then performed USG and MRI but only diagnosed with hypoplastic FPL intraoperatively.^{3,6}

A hypoplastic sized thumb compared to unaffected thumb may be a diagnostic feature of the absence or hypoplastic FPL, but in a bilateral abnormality, such as in our case, both thumb are of the same size. In a bilateral inability of IP joint flexion, the absent of IP joint crease (Fig 3) is a better diagnostic feature of absence or hypoplastic FPL.

Intraoperatively, we found that the FPL was hypoplastic and we could perform weaving with FDS IV with Pulvertaft technique. In cases with the absence of FPL, restoration of IP flexion can still be performed with the FDS IV fixed transosseously through the base of the distal phalanx of the thumb.⁴

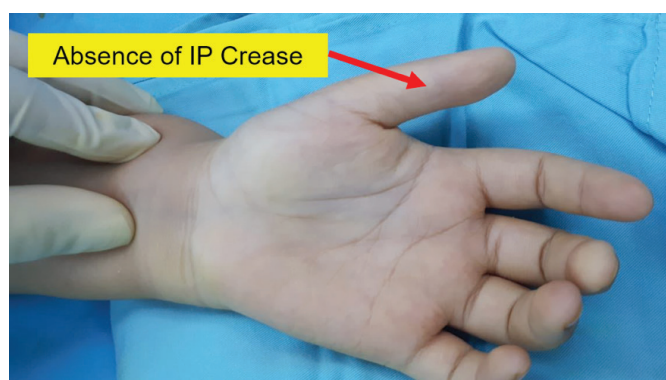


Figure 3. Inability of IP joint flexion, the absent of IP joint crease as a diagnostic feature of the absence or hypoplastic FPL

CONCLUSION

We concluded that the absence of interphalangeal crease during physical examination is one of the obvious signs of FPL absence or hypoplasia. The use of dorsal capsule release and tendon transfer in treating FPL absence or hypoplasia without any other thenar anomalies is proven to be adequate and feasible.

Conflict of interest

None to be declared

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