

## Congenital bilateral absence of flexor pollicis longus tendon without associated anomalies of thumb hypoplasia: A case report

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### ABSTRACT

The human thumb is the most important integral part of our hand because of its unique pattern of development which makes it the most unique digit of the hand. Any deviation from the normal development of the thumb leads to serious malfunction and cosmetic problems. One of the rarest types of anomaly is the congenital bilateral absence of flexor pollicis longus (FPL) tendon without thumb hypoplasia and thenar atrophy. In this paper, we are reporting the case of a 30-year-old girl with the congenital bilateral absence of FPL tendon without any associated anomalies of the thumb hypoplasia. The diagnosis is confirmed if the patient is not able to flex the interphalangeal joint of both the thumbs in the presence of an absence of interphalangeal joint creases. Apart from physical examination, we have also used radiographic and magnetic resonance imaging to diagnose our case.

**Key words:** *Flexor pollicis longus tendon hypoplasia, Hand, Thumb hypoplasia*

The human thumb is the most important integral part of our hand because of its unique pattern of development which makes it the most unique digit of the hand. It lies in the plane of palm and perpendicular to all other digits. It is prehensile and opposable. It is responsible for pinch, grasp, fine motor skills, and dexterity. Due to this reason, any deviation from the normal development of the thumb leads to serious malfunction and cosmetic problems [1]. One of the rarest types of anomaly is the congenital bilateral absence of flexor pollicis longus (FPL) tendon without thumb hypoplasia and thenar atrophy [2-5]. In 1979, Arminio was the first to report the congenital absence of FPL tendon. Since then, many such cases have been published till date. Most of those published cases are unilateral absence of FPL tendon. Bilateral absence is extremely rare. Very few cases of congenital bilateral absence of FPL tendon have been reported so far, in which the last case had additional findings of bilateral thumb hypoplasia with thenar atrophy. The true prevalence of such cases is still unclear.

Here, we are reporting a case of congenital bilateral absence of FPL tendon without thumb hypoplasia and thenar atrophy in a 30-year-old girl. The patient came to us seeking some other medical attention and the diagnosis was made by chance because it was not her main concern as she was well adjusted to her day-to-day life without any functional deficit.

### CASE REPORT

A 30-year-old woman presented to our hospital outpatient department with chief complaints of difficulties in performing activities such as holding an object within the compass of both hands and writing,

although she had no major difficulties while performing her daily routine activities. She had been unable to flex her both thumbs at the interphalangeal joint. She was not aware of this deficit which was present in both the thumbs until she was in her pre-school as she could not able to write properly. There was no history of any childhood injury or any similar complaints in other family members. There was no family history of any birth defect as well.

Her general physical examinations were normal and the vitals were stable. Local examination shows minor hypoplasia of both thumbs and an absence of dorsal wrinkles (Fig. 1a) and flexion creases (Fig. 1b) at the interphalangeal joints of both the thumbs. There was no loss of normal muscle contour of thenar eminence bilaterally (Fig. 1b). On performing a range of motions, active flexion at the interphalangeal joints was absent bilaterally although 30° of interphalangeal joint flexion was present on passive motion bilaterally. Bilateral thumb opposition was also weak. Pinch strength was also reduced bilaterally and equal in amount.

The radiograph of the hands showed no skeletal changes (Fig. 2). Magnetic resonance imaging (MRI) of both hands showed the absence of FPL muscle bilaterally (Figs. 3 and 4). At the end of our clinical and radiological examinations, we have concluded that it is a case of congenital bilateral absence of FPL tendon without associated anomaly of thumb hypoplasia. The patient was managed conservatively.

### DISCUSSION

Inability to flex the interphalangeal joint of thumbs can be due to multiple causes, but congenitally inability to flex the interphalangeal

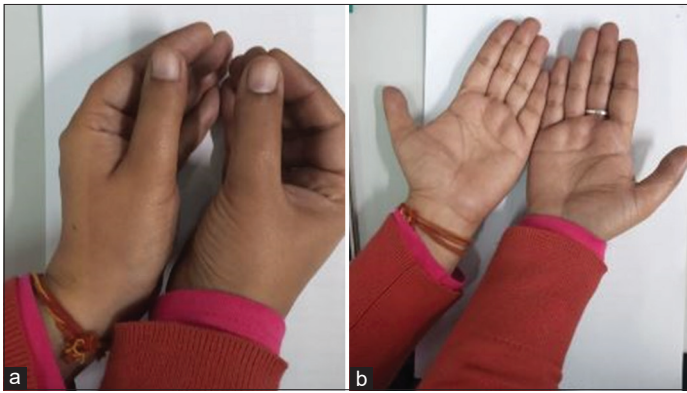


Figure 1: (a) Dorsal view of bilateral thumb showing absence of dorsal wrinkles; (b) absence of flexion creases over both thumbs



Figure 2: Radiograph shows no skeletal hypoplasia

joint of thumbs may be due to congenital absence of FPL, anomalous insertion of FPL, congenital tenovaginitis of FPL, or partial anterior interosseous nerve palsy. Among all, the congenital absence of FPL is an extremely rare cause. Thumb hypoplasia and thenar muscle atrophy are also associated with congenital anomaly of FPL but without thumb hypoplasia and thenar muscle atrophy, FPL anomaly is rarest [6-9]. Congenital absence of thenar muscle (e.g., flexor pollicis brevis and abductor pollicis brevis) without the absence of FPL has also been reported in the literature [1]. FPL deficiency is usually unilateral but rarely bilateral. Most cases are reported in pediatric age groups only a few are reported in adults.

Clinically, the affected thumbs have a loss of dorsal wrinkles and flexion creases. There is also a loss of active flexion on the interphalangeal joints of the thumb. Other findings such as soft tissue and skeletal hypoplasia may or may not be associated. Blauth *et al.* classified thumb hypoplasia as isolated minor hypoplasia (type 1); associated with thenar hypoplasia and metacarpophalangeal (MCP) joint instability (type 2); musculotendinous/osseous deficiency with absent active motion at MCP or interphalangeal joint (type 3); floating thumb (type 4); and complete absence of thumb (type 5).

Along with clinical examination, radiography, MRI, and electromyography can be used for diagnostic purposes, documentation, and surgical planning. Real-time ultrasonography (USG) may help to evaluate the dynamicity of the tendons. USG has an advantage over other modalities that there is no radiation



Figure 3: T1-weighted coronal image of hand shows absence of flexor pollicis longus tendon

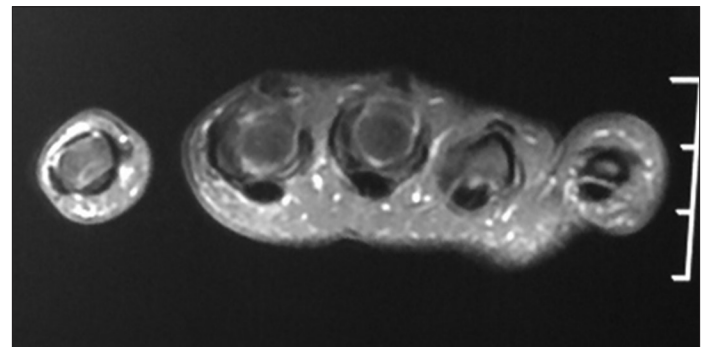


Figure 4: Axial image of the hand shows absence of flexor pollicis longus tendon

exposure to the patients and it carries a low cost, safe, and easy to perform, but MRI is highly accurate in the diagnosis over other modalities [10].

To achieve flexion at the interphalangeal joint of the thumb, the most preferred surgical technique is tendon transfer followed by rigorous rehabilitation. The surgical reconstructive technique is either one staged or two staged using flexor digitorum superficialis tendon of the ring finger. The achieved range of flexion after the one-staged operation was similar to the achieved range of flexion after the two-staged operations and varies between 20 and 35° in both groups. Following surgery, vigorous rehabilitation in the form of physiotherapy is must to obtain satisfactory results [3,8].

In our case, the patient was managed conservatively and counseled as patient and her parents are not keen for any surgical interference. Although they have been evaluated for the benefits of surgery, they believed that she has already adapted in her all activities of daily living without any limitations. The patient was advised for regular follow-up and follow-up when needed. Thomas and Mathivanan had reported a similar case in 1999 [8].

## CONCLUSION

Most cases are reported in the pediatric age groups and only a few are reported in adults. Hence, surgical intervention is not always

a definitive treatment of such anomaly if the presentation of such cases appeared late in life where the patient has well adapted in her day-to-day activity including fine grasp, writing, and typing without any limitations.

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