Advanced Hypoplasia of the Flexor Pollicis Longus Tendon in Twins

İkizlerde Fleksör Pollisis Longus Tendonunun İleri Derecede Hipoplazisi

Flexor Pollicis Longus Hipoplazisi / Hypoplasia of Flexor Pollicis Longus

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Özet

Fleksör pollisis longus (FPL) tendonunun ileri derecede hipoplazisi literatürde nadir karşılaşılan bir anomalidir. Bu çalışmada, 2.5 yaşında ikiz kardeşler sunuldu. İlginç olarak, her iki kardeşte FPL tendonunun belirgin hipoplazisi saptandı ve eşlik eden başka bir anomali yoktu. Fizik muayene ve manyetik rezonans görüntüleme ile birlikte olgular incelendiğinde FPL tendonunda hipoplazi saptandı.

Anahtar Kelimeler

Fleksör Pollisis Longus; Tendon; MRG; Hipoplazi; İkiz

Abstract

Advanced hypoplasia of the flexor pollicis longus (FPL) tendon is a rarely encountered anomaly in the literature. 2.5-year-old twins were reported in this study. Interestingly, marked hypoplasia of the FPL tendon was detected in both twins, and no other accompanying anomaly was present. Hypoplasia of the FPL tendon was detected when the cases were subjected to physical examination and magnetic resonance imaging.

Keywords

Flexor Pollicis Longus; Tendon; MRI; Hypoplasia; Twins

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Introduction

Congenital anomalies of the thumb create more serious functional problems than the anomalies of the other digits. The congenital absence of the flexor pollicislongus (FPL) tendon is rarely reported in the literature [1,2]. The absence of dorsal interphalangeal crease can be used as a diagnostic tool for some cases. Only nine cases of tendon absence have been reported in the literature (Table).

Table: A list of the cases with congenital absence and hypoplasia of the FPL tendon in the literature

Case No	Sex	Age	Diagnosis	Reference
1	Female	1	PE	Miura 1977
2	Female	6	PE	Arminio 1979
3	Female	13	PE, EMG	Koster 1984
4	Female	7	PE, EMG	Uchida et al. 1985
5	Male	5	PE	DeHaan et al. 1987
6	Female	11	PE	Thomas and Mathivanan 1999
7 (hypoplasia)	Male	10	MR	Alicioglu 2007
8	Female	14	X ray, MR	Demirseren et al. 2007
9	Male	43		Lutes and Tamurian 2007
10	Male	28	X ray, CT	Sures 2009
11 (hypoplasia)	**Male**	**2.5**	**PE, X ray, MR**	**Present case**
12 (hypoplasia)	**Male**	**2.5**	**PE, X ray, MR**	**Present case**

Advanced hypoplasia of the FPL tendon is rarely reported in the literature. We report 2.5-year-old twin cases in this study. Interestingly, these cases are twins with marked hypoplasia of the FPL tendon but no other accompanying anomaly.

Case Report

Two-and-a-half-year-old right-handed male twins were presented with the inability to flex their right and left thumbs at the interphalangeal joint. The patients were subjected to a thorough physical examination, X-rays, and the magnetic resonance imaging (MRI) of the hands, and hypoplasia of theFPL tendons was detected.

There was neither family history of congenital anomaly nor any trauma. They had no additional abnormality in the physical examination. Upon the physical examination, both thumbs were observed to be hypoplastic. However, no thenar atrophy existed. The dorsal wrinkles were less evident at the interphalangeal joint of the thumb of both hands (Figures 1, 2, and 3). There was no active flexion at the interphalangeal joints. The passive range for the motion of the thumbs was restricted on both

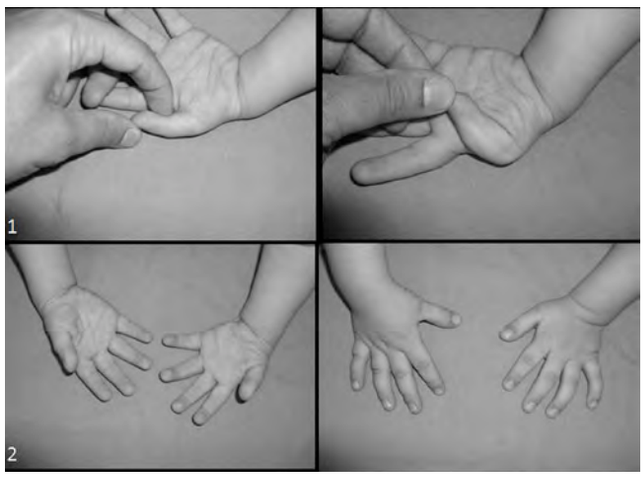


Figure 1. Absence of dorsal wrinkles on the thumb was detected
Figure 2. The findings are followed in both hands of the twins

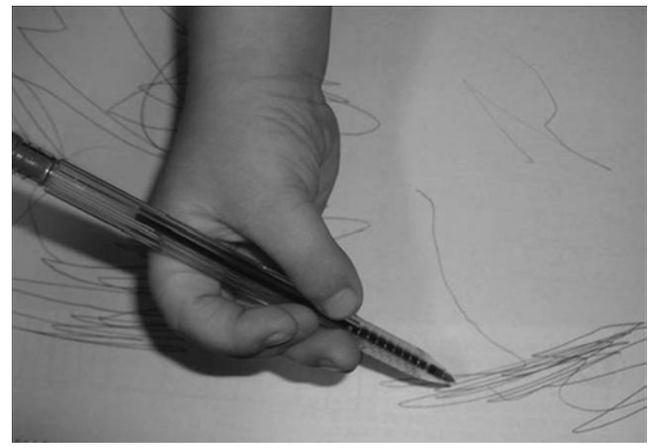


Figure 3. There was no dorsal wrinkle on the thumb

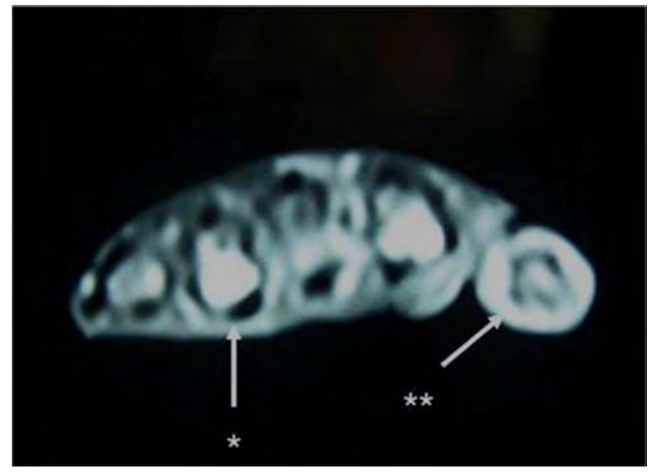


Figure 4. Hypoplasia of the FPL tendon was shown at the transverse T1W section (**) Normal tendon (*)

hands.Thenar eminences were similar in size. The movements of the metacarpophalangeal and carpometacarpal joints were normal in the physical examination. The X-rays of the hands did not show any hypoplasia of the first phalanges. We did not perform any ultrasonographic examination, for the patients were very young. The MR images of both hands showed that the FPL tendon was in its normal location and extremely thin (Figure 4). This finding and the physical examination finding led to the diagnosis of the hypoplastic tendon.

Discussion

To date, there have been no reported cases of twins with the hypoplasia of FPL tendon in the literature. The diagnosis should be kept in mind for any patient with a hypoplastic thumb who is unable to flex the interphalangeal joint. The congenital inability to flex the interphalangeal joint of the thumb may be due to one of the several causes, including congenital tenovaginitis of the flexor tendon sheath, and partial anterior interosseous nerve paralysis [2-5].

Congenital anomalies of the FPL show various anatomical features. They are classified according to whether there are other associated anomalies, the severity of anomalies, and the affected site of the tendon [3-5]. It can be further divided into four groups as absence, abnormal insertion, abnormal course, and connection.

FPL absence is frequently associated with thenar muscle hypo-

plasia. Thenar muscle hypoplasia is understandable; however,the cause of absence of thenar atrophy is mysterious [2-5]. Aplasia or hypoplasia of the FPL tendon is generally isolated; nevertheless, some cases are accompanied by other congenital anomalies.

Usami reported bilateral FPL tendon absence with craniofacial anomalies [6]. Skeletal hypoplasia of the thumb has been reported in a case report [1,3,4,7]. However, our cases had no skeletal hypoplasia.

Alicioğlu [3] reported a 10-year-old boy with the inability to flex the right thumb. In the report concerned, the patient stated that the problem had been present for two months. There was no history of injury or other congenital anomaly. Besides, regarding that case, it was stated that no thenar atrophy existed, and both hands were observed to be symmetrical in the physical examination. Our cases were very young in comparison with that case; both thumbs were observed to be hypoplastic in the physical examination; and there was no thenar atrophy.

FPL tendon abnormalities are commonly isolated; however, some cases are related to congenital anomalies. In 1988, Hagan and Idler demonstrated that a patient with abnormal insertion of the FPL tendon had surgical correction of a congenital ureteric anomaly [8] . Nevertheless, no evidence of the genetic background of this condition was available.

The current report presents a unique finding about the hypoplasia of FPL since it has never been encountered in twins with the involvement of both hands.

Ultrasonography (US) is a considerably cheap and easy technique for diagnosis. However, it is difficult to obtain a good result in very young children by means ofthe US due to the lack of cooperation. There was a ten-year-old patient who was diagnosed by means of US in the literature [3]. Our cases were 2.5 years of age, and no appropriate US probe was available for US examination. In addition, the disadvantages of US imaging include operator dependence, the difficulty in providing the detailed knowledge of anatomy, and understanding of the potential pitfalls. Therefore, MRI can also be highly accurate and should be considered a diagnostic technique for those patients in the case of whom US cannot be used.

As the surgical treatment depends on important anatomical landmarks, the authors elucidated the feasibility of MRI in the evaluation of flexor tendon morphology of the hand. MRI is not only anaccurate diagnostic technique but also beneficial to the treatment procedure. Furthermore, MRI is a highly sensitive technique to demonstrate tendon pathologies.

In conclusion, genetic transfer seems to be a major cause of the hypoplasia of the FPL tendon. Thus, the role of inheritance during the development of advanced hypoplasia of the FPL tendon is yet to be defined. Early diagnosis and supportive treatment will enable to easily overcome all problems that may be caused by the anomaly.

Competing interests

The authors declare that they have no competing interests.

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