Flexor Pollicis Longus Hipoplazisi / Hypoplasia of Flexor Pollicis Longus

İkizlerde Fleksör Pollisis Longus Tendonunun İleri Derecede Hipoplazisi

Fleksör polisis longus (FPL) tendonunun ileri derecede hipoplazisi literatürde nadir karşılaşılan bir anomalidir. Bu çalışmadada, 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 bu iki oğulun incelendikinde FPL tendonunda hipoplazisi saptanıldı.

Anahtar Kelimeler
Flexor Pollicis Longus; Tendon; MRG; Hipoplazia; İkiz

Özet
Flexor polisis longus (FPL) tendonunun ileri derecede hipoplazisi literatürde nadir karşılaşılan bir anomalidir. Bu çalışmadada, 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 bu iki oğulun incelendikinde FPL tendonunda hipoplazisi saptanıldı.

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
Introduction
Congenital anomalies of the thumb create more serious functional problems than the anomalies of the other digits. The congenital absence of the flexor pollicis longus (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).

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 the FPL 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 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 hypoplasia. The th
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.

Alicioglu [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 an 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.

Ultrasoundography (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 of the 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 an accurate 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.

References

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