Observation of the Improvement in the Active Range of Motion by a Short-Term Active Thumb Flexion Exercise Is Helpful in the Diagnosis of Flexion Disturbance in Bilateral Thumbs Due to Anomalous Tendon Slip of Flexor Pollicis Longus - Congenital Flexion Disturbance in Bilateral Thumbs: a Case Report

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We report an 11-year-old boy with limitation of flexion in both thumbs due to anomalous tendon slips of the flexor pollicis longus (FPL). The patient had been unaware of inability to flex his thumbs until his teacher noticed difficulty in writing. Since there were no flexion creases at the interphalangeal joint on either of his thumbs, severe congenital anomalies such as the absence of thumb flexion force and other disorders were suspected. However, because his active range of motion was improved with a short-term self-active flexion exercise program, he was diagnosed as having congenital anomalous tendons with FPL insertion sites other than the tendons of his other fingers. Surgical division of the anomalous tendon slips alone yielded a good functional outcome.

Key words: anomalous tendon slip, flexor pollicis longus, congenital anomaly

Introduction

Non-traumatic impairment of thumb flexion can be induced by various factors. Linburg and Comstock reported anomalous tendon connection from the flexor pollicis longus (FPL) to the index finger flexor digitorum profundus in 1979. An anomalous tendon from the FPL to the flexor indicis profundus limited independent flexion of the thumb. When there is an anomalous tendon connection from the FPL to the tendon of another finger as reported by Linburg and Comstock, that finger moves in conjunction with flexion of the thumb, and this facilitates establishing the clinical diagnosis. In our patient, however, the anomalous tendon slips from the FPL had mainly inserted in the flexor retinaculum bilaterally. This hampered the flexion of both thumbs, and no flexion of the other fingers accompanying thumb flexion was observed. Thus, we prescribed thumb self-active range of motion (ROM) exercises for this patient to observe whether active ROM would improve. As a result, active ROM of both thumbs improved, and we were thus able to confirm the diagnosis of a congenital anomaly, i.e. anomalous tendons of the FPL were inserted in sites other than the tendons of the other fingers.

Case Report

An 11-year-old Japanese boy complained of inability to flex both of his thumbs at the interphalangeal (IP) joint. His teacher was the first to observe that he had difficulties with writing and drawing, but the patient and his parents had been unaware of his inability to flex his thumbs because he had performed most tasks with both hands without noticeable difficulty. There was no family history of congenital anomalies. The thumbs and thenar eminences of both hands were essentially the same size. There were no flexion creases at the interphalangeal joint on either of his thumbs, severe congenital anomalies such as the absence of thumb flexion force and other disorders were suspected. However, because his active range of motion was improved with a short-term self-active flexion exercise program, he was diagnosed as having congenital anomalous tendons with FPL insertion sites other than the tendons of his other fingers. Surgical division of the anomalous tendon slips alone yielded a good functional outcome.
Figure 1 Photographs of the (A) left thumb and (B) right thumb. No flexion creases can be seen at the IP joint on either side.

Figure 2 A. At the first visit to our hospital. At presentation, active flexion of the left thumb at the IP joint was possible with a range of 0/15 degrees. B. After performing an active thumb flexion exercise program for 2 months, active ROM increased to 0/30 degrees on the left side. C. At the first visit to our hospital. At presentation, flexion of the right thumb at the IP joint was not possible. D. After performing an active thumb flexion exercise program for 2 months, flexion of the right thumb at the IP joint was still not possible.

Figure 3 Radiographs of the posteroanterior (A) and lateral (B) views of the left thumb, and posteroanterior (C) and lateral (D) views of the right thumb.
creases at the IP joint on either side (Figure-1A, B). The IP joint of the right thumb remained extended at all times, but active flexion of the left thumb at the IP joint was possible with a range of 0/15 degrees (Figure-2A, C). Passive ROM of the IP joints was 0/30 degrees on both sides. The ROM and sensibilities of all other digits were normal.

Radiographs of the thumbs showed that the joint surface of the proximal phalange at the IP joint was slightly flattened in both thumbs (Figure-3A, B, C, D). Magnetic resonance imaging (MRI) of the thumbs revealed articular cartilage of normal thickness in the thumb IP joint lining both the proximal and distal phalanges, and the FPL tendons were normal in both thumbs (Figure-4).

Since the FPL tendon and articular cartilage appeared normal in both thumbs, we considered the possibility of congenital anomalies in which anomalous FPL tendons were inserted into sites other than the tendons of the other fingers, and initially prescribed an active ROM exercise program for two months. Active ROM increased from 0/15 to 0/30 degrees on the left side (Figure-2B, D), and flexion creases were also ultimately established at the thumb IP joint. Because the active ROM was still inadequate and given the above-mentioned findings, we suspected that there were anomalous tendon slips of the FPL which inserted into sites other than the tendons of the other fingers. Surgery was first performed on the left side, and 2 months later on the right side.

During the operation on the left side, a zig-zag palmar incision was made and the flexor retinacu-

lum was explored from the distal to the proximal end. Several well developed anomalous slips from the original FPL were found inserting into the flexor retinaculum and palmar fascia through the carpal tunnel (Figure-5). Division of the anomalous slips allowed full extension of the FPL, and side-to-side suturing of the slips to the original FPL tendon was performed in order to maintain the dynamic strength and power of the tendon without reducing its output power as the source. Two months later, we performed a similar operation on the right side. Several well developed anomalous slips from the original FPL were found to be inserted into the radial wall of the carpal tunnel and the flexor retinaculum (Figure-6). Division of the
anomalous slips allowed full extension of the FPL, and a side-to-side suturing of the slips to the original FPL tendon was performed, for the same reasons as described above for the left side. He achieved active flexion of 60 degrees on the left side at eight months postoperatively and of 45 degrees on the right side at six months postoperatively. Although the patient had not been aware of any serious difficulties in his daily life even before the surgery, he and his family are satisfied with the improvement in the active ROM of both thumbs.

Discussion

Thumb flexion limitations in a child can have many possible causes. The causative factors reported in the literature to date include congenital absence of the FPL, congenital trigger thumb, radial ray hypoplasia, old trauma, peripheral nerve palsy, and anomalous insertion of the FPL tendon. Therefore, making an exact clinical diagnosis can be difficult. In our case, MRI was useful for the differential diagnosis because the FPL tendon and articular cartilage were clearly identified in both thumbs. Based on these findings, the diagnosis of congenital absence of the FPL tendon was excluded, but the possibility of congenital absence of the flexion force of the FPL could not be ruled out because there were no flexion creases on the volar side of the thumb IP joint. However, the short-term thumb active ROM exercise program performed by the patient himself improved his active flexion ROM, and surprisingly, even flexion creases were ultimately established. These observations suggest that despite the active flexion of his thumbs being limited for various reasons, the FPL tendons did exist and were inserted into the distal phalanges beyond the IP joints, under conditions in which muscle exercises are effective for improving function. The patient was able to flex his thumb IP joint when he intended to do so, and active ROM improved with exercises though full recovery was not achieved. These improvements would not have occurred had his condition been simple hypoplasia of the tendon. These observations indicated that the patient had a congenital anomaly, i.e. anomalous tendons of the FPL inserted into sites other than the tendons of his other fingers. Anomalous tendon insertion of the FPL occurs in two forms: unusual attachment of the FPL tendon and anomalous tendon slip to flexor digitorum profundus II. Our patient had the former type, and since his other fingers did not flex in conjunction with the flexion of his thumbs, it was difficult to obtain an accurate clinical diagnosis. The short-term thumb active ROM exercise program performed by the patient himself was remarkably helpful in establishing the diagnosis.

The patient and his parents had been unaware of the patient’s inability to flex both of his thumbs until his teacher noticed that he had difficulties with writing and drawing. A possible reason is that the anomaly was bilateral and did not cause significant inconveniences in his routine daily activities. Linburg and Comstock reported that 12 of 48 cadavers (25%) examined had anomalous tendon slips from the FPL in one extremity and three specimens (6%) had the anomaly bilaterally. Several cases of limited thumb flexion due to insertion of an anomalous slip of the FPL have previously been reported. In our case, flexion was more markedly improved in the left thumb than in the right thumb. Inability to regain full flexion of the right thumb after operative release may have resulted from the relatively poor volume of his weaker FPL tendon in the right thumb as compared to the left thumb. In this patient, surgical division of the anomalous tendon slips alone yielded good functional outcomes, for both thumbs, without additional tendon transfer or other procedures.

References