Case Report

Flexor carpi radialis brevis muscle, a rare anomalous muscle of the forearm; a case report and literature review

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Abstract: The flexor carpi radialis brevis (FCRB) is a rare abnormal muscle of the distal forearm or wrist. Its incidence varies in different reports, which oscillating between 2-8%. This paper reports a case of FCRB found in the anatomy of the forearm. The abnormal muscle, which started from the facies volaris distal radii, occupies the terminus of pronator quadratus, and was observed with dysplasia in the pronator quadratus. We also reviewed the literature on FCRB anatomy, especially the surgical exposure of distal radius fractures and the clinical symptoms caused by FCRB. Knowledge of the anomalous muscles of the forearm and wrist can highly improve clinician's understanding of the disease during surgery and imaging examination, and reduce unnecessary trauma caused by misdiagnosis.

Keywords: Flexor carpi radialis brevis (FCRB), anomalous muscle, forearm

Introduction

Flexor carpi radialis brevis (FCRB), among the anomalous muscles of the forearm and wrist, is one of the extremely rare anomalous muscles. In 1851, Fano [1] first reported the flexor carpi radialis as an anomalous muscle of the forearm, and called it "Radiocarpien". It was further described by Woods [2] in 1867 and the term "flexor radial" was created. In 1897, Le Double [3] classified FCRB variants into four types according to different morphologies of the muscle belly and tendon configuration, including the Radio-Palmaire type, the Radio-Carpien type, the Radio-Metacarpien type, and the Mixed type, of which the second was the most common. According to related literature reports, the occurrence rate of FCRB appears to be 2.6% in the Japanese population [4], 4-7.5% in European white subjects [3], and 3.4% in African American subjects [5]. As the anomalous muscle rarely causes clinical symptoms, it is often overlooked in imaging examinations of the forearm and wrist. Because of this, it is only occasionally reported in autopsies and surgical exploration. In this report, we provided a case study of a dual-forearm FCRB and a retrospective analysis of the existing published literature on FCRB.

Case report

During the dissection of the anterior group muscles in a left forearm specimen, we found a spindle-shaped penniform muscle in the deep side of the flexor digitorum superficialis (FDS) and flexor pollicis longus (FPL). The muscle started from the facies volaris distal radii and moved along the radialis of the flexor pollicis longus. The anterolateral side of the radius. which is at the level of the processus styloideus radii plane, is the muscle-belly of the muscle, and it occupies the terminus of pronator quadratus. According to the examination, the pronator quadrates were found with dysplasia, and was moved and changed to the tendons around the level of the radiocarpal joint. The tendon traveled along the ulnar side of the flexor carpi radialis (FCR) tendon, passing through the carpal canal together, and ending at the base of the palmar side on the second metacarpal bone (Figure 1). When the tendon was stretched, wrist flexion occurred. The blood supply came from a branch of the radial artery, and was

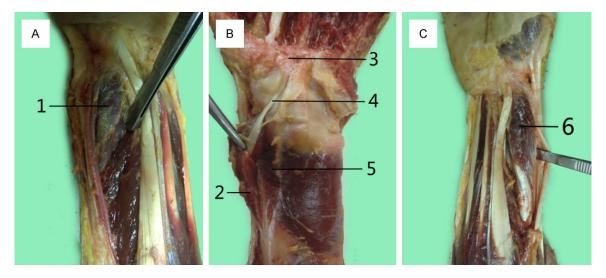


Figure 1. Left forearm: The muscle started from the facies volaris distal radii and ending at the base of the palmar side of the second metacarpal bone. 1: Flexor carpi radialis brevis (FCRB); 2: The muscle-belly of the FCRB; 3: Base of the second metacarpal bone; 4: The tendon of FCRB; 5: The insertion of the pronator quadratus in the radius (dysplasia in pronator quadrates).

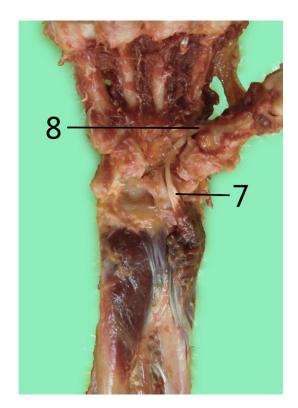


Figure 2. Right forearm: 6: Flexor carpi radialis brevis (FCRB); 7: The tendon of FCRB; 8: The insertion of FCRB is at the base of the first metacarpal base.

innervated by the anterior interosseous nerve. No other abnormal muscles or bone structures were found in the dissection of the specimen. Besides, the same situation was observed dur-

ing dissection of the subject's right forearm (**Figure 2**). However, the insertion of FCRB was at the base of the first metacarpal base.

In addition, we searched the literature published in PubMed, EMBASE, and Ovid databases up to December 2018 by using "Flexor carpi radialis brevis" and "FCRB" as keywords. A total of 17 full-text articles were retrieved, including 27 cases of patients with FCRB (Table 1).

Discussion

FCRB is a rare abnormal muscle of the distal forearm, and its incidence varies in different reports, which fluctuates between 2-8%. Among the total incidence of FCRB, the rate of FCRB was 4-7.5% in Europeans [2], 1.8-2.6% in Japanese, and 1.9% in Japanese autopsies [8], and 3.4% in African Americans [21]. FCRB often originates from the volar side of the lower third of the shaft of the radius, and the area between the origin of the flexor pollicis longus and the end of the pronator quadratus at the radius [6, 7, 16]. It travels on the ulnar side of the flexor carpi radialis, stretching across the surface of the pronator quadratus, and changes into the tendon at the level of the radiocarpal joint surface. It passes through the radial carpal canal parallel to the tendon of the radial flexor carpal muscle and enters the metacarpus. However, there are also reports [20] where the FCRB tendon crosses the FCR ten-

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 Table 1. Literature study on Flexor carpi radialis brevis

Author	Years	Case	FCRB originates	FCRB insertion	FCRB subtype	Function	Hypoplasia of the PQ	Cause
Dodds [6]	2006	1	Distance from radial styloid process 6 cm	The base of the second metacarpal bone, trapezial	Muscle-belly	Wrist flexion	YES	Cadaveric dissection
Nagata et al. [7]	2016	1	Lower one-third of the radius	-	Muscle-belly	-	YES	Distal radius fractures
		1	Lower one-third of the radius	-	Muscle-belly	-	YES	Distal radius fractures
		1	Lower one-third of the radius	-	Muscle-belly	-	NO	Distal radius fractures
		2	-	-	Muscle-tendon		NO	Distal radius fractures
Nakahashi and Izumi [8]	1987	1	Occupied the distal region of the flexor pollicis longus origination	The base of the second, third metacarpal bone	Muscle-tendon	-	YES	Cadaveric dissection
		1		The third metacarpal bone	-	-	-	Cadaveric dissection
Mantovani et al. [9]	2010	1	-	-	Muscle-tendon	-	-	Distal radius fractures
Chong et al. [10]	2009	1	-	-	-	-	-	Hand injury
Duncan et al. [11]	2006	1	The radius	The base of the second metacarpal bone	-	-	-	MRI
Smith and Kakar [12]	2014	1	-	-	-	-	-	Ultrasound and MRI
Peers and Kaplan [13]	2008	1	Lower one-third of the radius	Trapezium	Muscle-tendon	-	YES	Painful mass on the radial
Mimura et al. [14]	2017	1	The anteroradial aspect of the volar radius	The base of the second metacarpal bone	Muscle-tendon	Wrist flexion	YES	Wrist pain
Hongsmatip et al. [15]	2018	1	The radius	Trapezium	-	-	-	Intersection syndrome of the wrist
Lee et al. [16]	2014	1	-	-	Muscle-tendon	-	YES	Distal radius fractures
		1	Between the radial insertion of the PQ and the origin of the FPL	-	Muscle-tendon	-	YES	Distal radius fractures
Kordahi et al. [17]	2018	1	-	-	-	-	-	MRI
Urigo et al. [18]	2017	1	Between the radial insertion of the PQ and the origin of the FPL	Capitate, base of the second, fourth metacarpal bone	Muscle-tendon	-	YES	Painful mass on the wrist
Laugharne [19]	2010	1	-	The base of the third metacarpal bone	Muscle-tendon	-	-	Distal radius fractures
Kosiyatrakul et al. [20]	2010	1	-	-	-	Wrist flexion	-	Painful on the volar radial
Kang et al. [21]	2006	1	Lower one-third of the radius	-	Muscle-tendon	Wrist flexion	YES	Distal radius fractures
Ho et al. [22]	2011	1	The radius	-	Muscle-tendon	Wrist or fingers	NO	Distal radius fractures
		1	The radius	-	Muscle-tendon	flexion	NO	Distal radius fractures
		1	Originated from the radial side of the starting point of FPL	-	Muscle-tendon	-	-	Distal radius fractures
		1	Originated from the radial side of the starting point of FPL	-	Muscle-tendon	-	-	Distal radius fractures
		1	Distal radial side	Trapezium		-	-	Cadaveric dissection

don from the ulnar to the radial side. There is a variety of termini in FCRB tendon, including the trapezial, trapezoid and capitate bones, and the base of the second, third, and fourth metacarpal bones [6, 7, 11, 13-15, 18, 19]. The FCRB is innervated by a branch of the median nerve, the anterior interosseous nerve [6, 8, 14]; whereas the blood supply is provided by the anterior interosseous artery [8]. mediating the functions of wrist flexion and finger flexion [6, 14, 20-22]. Mimura et al. [14] described the occurrence of FCRB contraction when stimulating the anterior interosseous nerve with an electrical nerve stimulator during surgery. In this study, the autopsy of the forearm muscle groups revealed that the flexion of the wrist can be observed when FCRB tendon is extended by the anterior interosseous nerve innervation which was similar to the reports by Kosiyatrakul et al. [20] and Kang et al. [21]. In previous literature reports, FCRB was often accompanied with dysplasia of the pronator quadratus. Nagata J et al. [7] reported 5 patients with FCRB, in which 2 of 3 patients with muscle belly-type FCRB had pronator quadrates dysplasia, and the remaining 2 patients with tendon-type FCRBs were observed without PQ dysplasia. In this autopsy, the bilateral FCRB of the subject belonged to muscle-belly-type FCRBs and had the pronator quadratus dysplasia. Dodds et al. [6] believed that the muscle-belly of the muscle-belly type FCRB occupied the terminus of the pronator quadrates at the distal end of radius, resulting in the dysplasia of pronator quadratus.

With the increasing incidence of distal radius fractures in recent years, distal radius locking plate internal fixation with the Volar or Henry approach has been widely used by orthopedic surgeons, which has resulted in increased report rates of FCRB. After reviewing the relevant literature, we found 11 cases of FCRB were reported in the treatment of distal radius fractures, and 1 case was found in the treatment of wrist laceration [10]. Among them, Nagata et al. [7] reported 123 cases of distal radius fractures, and 5 of which was found with FCRB, with an incidence rate of 4.1%. Mantovani et al. [9] used locking plates with the Volar approach to treat 172 cases of distal radius fractures, and found 6 cases of FCRB, with an incidence of 3.5% (3 males and 3 females). Lee et al. [16] reported an incidence

of 2.8% in 71 cases of patients who underwent the same procedures and approaches. According to the experience of the above three authors, the placement of the locking plate would not be affected by the intraoperative detection of FCRB, and postoperative complications were seldom occurred. Only one case [7] reported pain at the radial side of the wrist joint 6 months after surgery, and the surgery was performed again. The synovial tissues around the FCRB tendon were observed with significant hyperplasia. Histological examination revealed that the synovial tissues were surrounded by small blood vessels with fibroblast hyperplasia and extensive inflammatory cell infiltration. This result indicated that FCRB tenosynovitis could possibly occur in FCRB patients with locking plate internal fixation.

At present, there are few reports on clinical symptoms caused by FCRB. Because the FCRB tendons are usually parallel to FCR tendons and pass through the bone fibers of the carpal canal to enter the wrist, it is generally unlikely to compress the median nerve to cause carpal tunnel syndrome. However, the FCRB can compress the anterior interosseous nerve, thus causing clinical symptoms [23]. A retrospective analysis of preoperative MRIs by Mimura et al. [14] in 379 patients with carpal tunnel syndrome revealed 6 cases of FCRB, with an incidence of approximately 1.6%. However, the causal relationship between FCRB and carpal tunnel syndrome remains unclear. Peers et al. [13], Hongsmatip et al. [15] and Kosiyatrakul et al. [20] have respectively reported the symptoms of cases caused by FCRB. All three reported patients had the same clinical symptoms: unexplained wrist joint pain and swelling at the radial side, which was aggravated during flexion of the wrist joint. Physical examination showed obvious pain upon pressure at the radial side of the wrist joint, a local mass, and negative Tinel's sign. Preoperative MRI and intraoperative examination found that the FC-RB tendon crossed the FCR tendon from the ulnar to the radial side. Hongsmatip et al. [15] believed this crossing could cause "intersection syndrome". In 1841, Velpeau described this disease for the first time. In 1987, Dobyns coined the term "intersection syndrome". which is also known as "crossover syndrome". The syndrome mainly occurred in the abductor pollicis longus, extensor pollicis brevis, and

between the tendons of the extensor carpi radialis longus and brevis muscles, and occasionally occurred in other abnormal crossing tendons [24]. A conservative approach has been recommended for the treatment of this disease, including physical therapy and nonsteroidal anti-inflammatory drugs (NSAIDs). For a very small number of stubborn cases or patients who needed quick relief from the painful symptoms, surgical incision should be conducted to the deep fascial region of the intersection region, and the possible bursae mucosae should be removed to limit the vicious cycle of muscle-tendon friction and swelling [25]. Two patients [13, 18] underwent intraoperative resection of the FCRB, achieving pain relief and normalization of wrist activity during follow-up. One patient [15] received physical therapy and NSAIDs for treatment, and the pain was ameliorated after 3 weeks. During operation, one patient [20] was found to have deformed FCRB tendons, and the gap between FCR tendons were filled with a large amount of synovial tissues. The gap was separated and resected during the operation, and the pain disappeared after 2 months of follow-up.

In conclusion, knowledge of the anomalous muscles of the forearm and wrist can highly improve clinician's understanding of the disease during surgery and imaging examination, and reduce unnecessary trauma caused by misdiagnosis. In addition to tenosynovitis, ganglion cyst and lipoma, anomalous muscle FCRB and intersection syndrome should be considered when pain, swelling and palpable mass were encountered in the radial side of wrist joint.

Disclosure of conflict of interest

None.

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