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A Fibroma of Tendon Sheath Causing Carpal Tunnel Syndrome: A Case Report of an Atypical Clinical Presentation 건수초섬유종에 의해 유발된 손목터널증후군: 비전형적인 임상 소견에 대한 증례 보고

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Case Report

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Fibroma of the tendon sheath is a benign slow-growing fibrous tumor. Although rare, cases occurring in the upper extremities usually involve the fingers. It appears as a well-defined, roundor oval-shaped mass originating from the flexor tendon. Abundant fibrous stroma makes fibromas appear as a low intensity mass in all MRI sequences. Most of the fibromas manifest as painless soft tissue masses. Herein, we report a case of fibroma of the tendon sheath with an unusual clinical presentation, triggering carpal tunnel syndrome during wrist movement.

Index terms Fibroma; Soft Tissue Neoplasms; Tendons; Carpal Tunnel Syndrome

INTRODUCTION

Fibroma of the tendon sheath (FTS) is a benign fibrous tumor with predilection for the flexor tendons (1). It manifests as a slow-growing, round or oval shaped mass in adults, usually in middle age male (2). Regarding its abundant fibrous component, FTS usually appear as low signal intensity at all sequences in MRI (2, 3). However, T2 signal intensity and the degree of enhancement can vary depending on its cellularity and internal myxoid change (3, 4). We experienced pathologically typical FTS showing unusual clinical presentation. In our case, FTS was symptomatic, moving dynamically in and out of carpal tunnel on wrist flexion and extension. Here, we report FTS with its imaging features related to its unusual clinical presentation.



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CASE REPORT

A 28-year-old female visited outpatient clinic of department of orthopedics, complaining a year-long right hand pain. She felt discomfort and rattling sensation during flexion and extension of the wrist. She also felt tingling sensation on right third and fourth fingers. On physical examination, there was a palpable mass deep in the palm. Ultrasonography (US) was done for further evaluation. She had no specific medial history. Laboratory tests were unremarkable.

On US exam, there was no significant abnormal lesion in carpal tunnel. There was no significant thickening of the flexor retinaculum. The median nerve in carpal tunnel showed normal shape and echogenicity. In palm, just distal to carpal tunnel, there was a 2 cm-sized ovoid hypoechoic mass along third flexor tendon. It was attached to flexor digitorum profundus tendon of third finger. On dynamic study, the mass slipped into distal carpal tunnel on flexion (Supplementary Video 1 in the online-only Data Supplement). On Doppler exam, there was no significant internal vascularity in the mass (Fig. 1A). MRI was recommended to characterize the mass.

MRI revealed 2.5 cm extent ovoid mass with partly ill-defined margin arising from flexor digitorum profundus tendon of third finger, just distal to distal carpal tunnel. The mass showed homogeneous isointensity on T1 weighted images, whereas on T2 weighted images, it showed heterogeneous variable intensity. After contrast injection, the mass showed hetero-geneous enhancement in peripheral portions. There was also mild enhancement along tendon sheath around the mass. Median nerve was intact (Fig. 1B). According to the incidence of soft tissue tumor by location, fibrous histiocytoma and tenosynovial giant cell tumor (GCT) are most common in wrist and hand, followed by FTS. However, the mass showed T2 high signal intensity, which is unusual for tenosynovial GCT and FTS. Furthermore, it had partly ill-defined margin which was suspected for infiltration. Thus, we thought fibrous histiocytoma would be more likely than tenosynovial GCT and FTS. We also mentioned the possibility of vascular tumor or venolymphatic malformation regarding heterogeneous peripheral enhancement and a small vessel abutting the mass.

The patient underwent excision of the mass. The mass appeared as whitish ovoid mass with indistinct margin, arising from tendon sheath of flexor digitorum profundus tendon of third finger (Fig. 1C). It was fibrotic mass and the mass slipped into carpal tunnel on flexion. The mass was removed whereas the tendon was preserved. Microscopically, the mass showed relatively high cellularity in central portion, whereas hypocellular with sparse numbers of bland spindled cells with hyalinized colloagenous stroma in peripheral portion. It was pathologically confirmed as FTS (Fig. 1D).

The study protocol conformed to the ethical guidelines of the 1975 Declaration of Helsinki.

DISCUSSION

FTS is uncommon which manifests as a well-defined slow-growing painless lesion in middle age adults (1). Most of FTSs occur in the upper extremities, predominantly the hand and wrist regions (4). Abundant fibrous component in FTS makes the lesion appear hypoechoic

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Fig. 1. A 23-year-old female with a fibroma, located deep in the palm, causing carpal tunnel syndrome. A. US reveals an approximately 1×2 -cm heterogeneously hypoechoic mass attached to the flexor digitorum profundus of the third finger, distal to the carpal tunnel. The mass slips into the distal carpal tunnel on flexion, triggering carpal tunnel syndrome. The mass shows no significant vascularity.

B. MRI reveals an approximately ovoid mass with partly ill-defined margin arising from the flexor digitorum profundus of the third finger, immediately distal to the distal carpal tunnel. On T1WI, the mass shows homogeneous isointensity. The mass shows heterogeneous variable intensity on T2WI, and no significant abnormality is seen in the adjacent median nerve (N). After contrast injection, the mass shows heterogeneous enhancement in the peripheral portions, with mild enhancement of the adjacent flexor tendons.

C. The excised specimen appears as a whitish, ovoid mass (M) arising from the origin of the third (*) and fourth flexor digitorum profundus.

D. Grossly the tumor is well circumscribed and lobulated. Microscopically, it shows relatively high cellularity in the central portion, whereas the peripheral portion is hypocellular with sparse numbers of bland spindle cells with hyalinized collagenous stroma. The cells have scant cytoplasm and elongated nuclei with evenly distributed fine chromatin (hematoxylin & eosin stain, \times 40).

AX = axial, CE = contrast enhanced, COR = coronal, fs = fat saturated, T1WI = T1-weighted image, T2WI = T2-weighted image, US = ultrasonography



in US (3). On MR, FTS typically shows low signal intensity on all sequences (2). In this case, unlike usual FTS, the mass showed a partly ill-defined border with heterogeneous high T2 signal intensity.

Soft tissue tumors arising from hand and wrist are usually benign (4). In relatively young aged adults, like our patient, fibrous histiocytoma is the most common, followed by tenosynovial GCT and FTS (5). Fibrous histiocytoma usually occurs within the dermis and subcuta-

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neous layer. This is so-called dermatofibroma which accompanies discolorization of overlying skin. It shows low to isointensity on T1WI and variable signal on T2WI. It also involves deeper soft tissues, but is rare compared to cutaneous counterpart (6, 7). Imaging features of FTS are similar to that of tenosynovial GCT, but there is some feature to distinguish these two entities. Hemosiderin deposition in GCT makes blooming artifact in T2WI, especially on gradient echo based sequences (8). Tenosynovial GCT shows strong enhancement after contrast administration because of its rich vascular proliferation, whereas FTS usually shows no or minimal enhancement (4). In our patient, the mass showed partly ill-defined margin which is unusual for FTS or tenosynovial GCT. We thought that indistinct margin can be infiltrative portion of deep seated fibrous histiocytoma. We also mentioned the possibility of vascular tumor or venolymphatic malformation, regarding a small vessel abutting the mass. Hemangioma is 4th most common soft tissue tumor in hand and wrist for young female. Though the mass did not show vascularity within the mass, hemangiomas without Doppler signal have been reported. We presumed small vessel abutting mass could contribute to peripheral enhancement. However, high signal intensity on T2WI is nonspecific, and there were no pathognomic findings of hemangiomas; vascular channels or fat overgrowth (6).

FTS can show variable T2 signal intensity and enhancement depending on its cellularity and myxoid change. T2 signal increases as its cellularity increases. Myxoid change also shows T2 high signal intensity (2). On pathologic exam, FTS in our patient showed more hyalinized collageneous stroma in peripheral portion. Its heterogeneity might cause heterogeneous T2 signal and enhancement. However, the mass manifested as a partly ill-defined mass on multimodality imaging. Similar to image findings, the mass had indistinct margin on operation. The surgeon resected the mass with part of adjacent lumbrical muscles. This is unusual for FTS because it usually shows as well defined mass (1). This atypical feature might be related with its dynamic movement. During flexion and extension, the mass moved dynamically in and out of the carpal tunnel. For a long time, this dynamic movement would cause wearing of the mass, resulting unusual feature.

There are few cases reporting unusual clinical presentation of FTS in wrist and hand (9, 10). These previously reported cases showed FTS causing trigger wrist or carpal tunnel. However, to our best knowledge, this is the first to focus on imaging features of FTS, related to its unusual presentation. In short, this is a case of FTS, moving dynamically during flexion and extension, resulting unusual clinical presentation.

In conclusion, FTS in hand and wrist usually manifests as asymptomatic benign mass. It is unusual, but the mass can cause symptoms on dynamic movement of the wrist, slipping with tendons. Dynamic movement can also cause local inflammation or associated wearing, leading to unusual imaging features. Radiologists should consider the incidence, imaging features and kinetics for proper diagnosis.

Supplementary Video Legend

Video 1. Longitudinal scan of proximal palm area. On wrist flexion, the mass slipped into distal carpal tunnel, causing median never compression.

Supplementary Materials

The online-only Data Supplement is available with this article at http://dx.doi.org/10.3348/



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Author Contributions

Conceptualization, K.A., L.G.Y., K.S.; data curation, all authors; investigation, all authors; methodology, all authors; project administration, K.A., L.G.Y.; resources, all authors; supervision, L.G.Y., L.J., H.H.W.; validation, all authors; visualization, all authors; writing—original draft, K.A., L.G.Y.; and writing—review & editing, all authors.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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건수초섬유종에 의해 유발된 손목터널증후군: 비전형적인 임상 소견에 대한 증례 보고

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힘줄의 섬유종은 천천히 자라는 양성 종양이다. 드물지만, 상지에 생길 수 있고 특히 손가락 굽힘 힘줄에 잘 생긴다. 섬유종은 대부분 경계가 좋은 원형 혹은 타원형의 종괴로 보이며, 풍 부한 섬유조직에 의해 자기공명영상의 모든 영상에서 어둡게 보이는 것이 특징이다. 대부분 의 섬유종은 무증상의 종괴로서 나타나지만, 드물게 증상을 야기하기도 한다. 우리는 손목의 반복적인 움직임에 의해 손목터널증후군을 유발하는 힘줄의 섬유종과, 그와 관련된 영상 소 견에 대해 보고하고자 한다.

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