Trigger wrist and carpal tunnel syndrome caused by hand intramuscular intrasynovial angiofibrolipoma: A rare case report

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ABSTRACT
Trigger wrist is a clinical entity characterized by triggering or the crackling of the wrist. Here, a case is reported of intrasynovial angiofibrolipoma that caused trigger wrist and carpal tunnel syndrome. This is the only case report where trigger wrist and carpal tunnel syndrome caused by the intrasynovial angiofibrolipoma were developed simultaneously. It is believed that that adhesive tenosynovitis developing in the tendons may have contributed to the triggering and carpal tunnel syndrome in the wrist as a result of inflammation occurring as a consequence of intrasynovial angiofibrolipoma.

Key words: Intrasynovial angiofibrolipoma, tenosynovitis, trigger wrist

Introduction
Trigger wrist is a clinical entity characterized by triggering or the crackling of the wrist. Suematsu et al. have identified the etiological factors that lead to these entities under three main categories: type A - tumor; type B - anomalous in muscle body; and type C - anomalous in tumor and muscle body [1]. The case reported here was categorized as type A. Trigger wrist and carpal tunnel syndrome caused by intrasynovial angiofibrolipoma in association with finger movements is a rare condition. In this report, a patient is described that has carpal tunnel syndrome developing in association with trigger wrist caused by intrasynovial angiofibrolipoma. Based on a review of the literature, this appears to be the first case report where trigger wrist and carpal tunnel syndrome brought about by intrasynovial angiofibrolipoma were developed simultaneously.

Case Report
A 40-year-old female patient was admitted to the authors’ Orthopedic Clinic with complaints of triggering of finger flexion and extension movements in the wrist and numbness in the 1st, 2nd and 3rd fingers in the hand. The complaints of the patient had been ongoing for approximately one year. No foreign bodies were found in the wrist or hand of the patient by way of inspection and palpation. Ultrasonography (USG), Magnetic Resonance Imaging (MRI) and Electroneuromyography (ENMG) tests were requested for the patient. Intrasynovial mass diagnosis was made in the palm area of the intramuscular region with USG and...
MRI (Figure 1). In ENMG, moderate sensory damage was also present in the area of the median nerve. Upon these diagnoses, it was decided that the patient required surgical intervention. After passing through the skin and subcutaneous tissue with an incision extending to the palm over the carpal ligament in the release surgery performed for carpal tunnel syndrome of the wrist, the carpal ligament was released and the median nerve was observed (Figure 2). After the median nerve was scru-
tinized, the incision was extended to the palmar and flexor profundus tendons. Flexor tendon inflammation was remarkable. Entering through the flexor tendons, the mass in the palmar region was excised together with synovium. Histopathological examination of the encapsulated tumor mass showed a composition of a large number of mature adipocytes, collagen fibrous tissue and blood vessels paved with variable-sized endothelial cells and positive immunoreactivity in vascular endothelial cells along with CD-34 primary antibody with no neoplastic elements (Figure 3). In the post-operative period, the wrist trigger along with the finger and wrist movements, as well as the numbness the patient felt in the median nerve tracing, disappeared.

**Discussion**

Triggering caused by flexor tendons in the wrist is a rare condition. It was first described by Eibel in 1961 [2]. When reviewing the literature, there are various instances of triggering and carpal tunnel syndrome caused by intrasynovial tumors in the wrist via the case reports [2-4]. Additionally, tumor-induced trigger wrist cases can also be found [5,6]. However, upon further review, no cases of triggering or carpal tunnel syndrome in the wrist caused by intrasynovial angiofibrolipoma could be found. As such, this is most likely the first case report where trigger wrist and carpal tunnel syndrome caused by the intrasynovial angiofibrolipoma were shown to be developed simultaneously. It is quite probable that adhesive tenosynovitis developing in the tendons may have contributed to the triggering and carpal tunnel syndrome in the wrist as a result of the inflammation caused by intrasynovial angiofibrolipoma.

Angiofibrolipoma is a scarce variant of lipoma that is a benign mesenchymal tumor and contains histopathologically mature adipocytes, blood vessels, and dense collagenous connective tissue. It bears macroscopic resemblance to lipoma [7,8]. An angiofibrolipoma is one of the rarest histopathological variants of a lipoma. Histological variants of lipomas contain fibrolipomas, angiolipomas, angiofibrolipomas, angiomyolipomas, and infiltrating angiolipomas [9]. Simple benign lipomas are the most common soft tissue tumor, but they are not desired in the hand. Fibroma of the tendon sheath are the predominant lesions with locations in the fingers,

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typically on the flexor surfaces, and they are most often seen in men. In a clinical manner, they grow stepwise, with expansion of the mass and occasional moderate tenderness. In addition, peripheral nerve sheath tumors (PNST) are seen in the palm region, clinically the reason for the common masses of the forearm and hand. In the hand and wrist, schwannomas originate from deeper and larger nerves and frequently develop along the flexor surfaces \([10,11]\). As is best presently known, intrasynovial angiofibrolipoma is exceptionally rare its causation of trigger wrist and carpal tunnel syndrome is unprecedented in the literature.

**Conflict of interest statement**

The authors have no conflicts of interest to declare.

**References**