Fibroma of the Tendon Sheath Arising from the Synovial Membrane of the Temporomandibular Joint

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Abstract: Fibroma of the tendon sheath (FTS) involving the temporomandibular joint (TMJ) is very rare. A case of FTS arising from the synovial membrane of the TMJ is presented. The patient was a 17-year-old male with jaw opening pain and clicking in the left TMJ. MRI, arthrotomography, and arthroscopy showed a well-circumscribed mass that was localized in the superior articular cavity of the left TMJ. The tumor was removed with the attached synovial membrane. Macroscopic, histological and immunohistological features of the intra-articular tumor were identical to FTS. There have been no signs of recurrence in the 6 years since surgery.

Key words: fibroma of tendon sheath, temporomandibular joint, intra-articular localization

Introduction
Fibroma of the tendon sheath (FTS) is a benign tumor that arises from the synovial membrane of the tendon sheath. The tumor consists of a slow-growing dense fibrous nodule that is firmly attached to the tendon sheath, and it frequently involves the fingers, hands, and wrists1. However, temporomandibular joint (TMJ) involvement by this tumor is extremely rare, and to our knowledge, only one case has been reported2. A case of histologically verified fibroma of the tendon sheath arising from the synovial membrane of the TMJ is presented.

Case Report
A 17-year-old male was referred to our hospital complaining of TMJ pain. He had had TMJ pain during opening and clicking in the left TMJ for 3 weeks. During mouth opening, the jaw deviated to the left side, and the maximum interincisal jaw opening was 39 mm. There was no swelling in the left TMJ region or malocclusion. He had no history of trauma or systemic joint diseases.

1. Imaging Findings
Magnetic resonance imaging (MRI) showed a mostly homogenous, well-circumscribed mass lateral to the lateral pterygoid muscle and immediately inferior to the articular eminence of the left TMJ. The mass demonstrated by MRI was visualized as a high signal intensity lesion on T2- and proton-weighted images and as an intermediate signal intensity lesion on T1-weighted images (Fig. 1A-C). Sagittal T1- and T2-weighted images showed the thickness of the synovial membrane as a hypointense curved line circumscribing the mass on the lateral portion of the left TMJ (Fig. 1C). Tomography showed superficial cortical bone erosion at the bottom of the articular eminence where it was in contact with the mass demonstrated by MRI (Fig. 2). Single contrast dual-space arthroto-
mography showed the presence of a mass lesion within the lateral portion of the anterior synovial pouch in the superior articular cavity (Fig. 3). Arthroscopy to the superior articular cavity of the left TMJ showed a grayish-white mass protruding into the anterior synovial pouch. However, fibrous adhesion or loose bodies were not seen in the superior articular cavity.

2. Surgical Findings

Based on clinical and imaging findings, a preliminary diagnosis of benign soft tissue tumor in the left TMJ was made. Under general anesthesia, an Al-Kayat & Bramy incision\(^3\) was made to approach the mass lesion. After the zygomatic bone periosteum was cut to open the superior articular cavity, a pedunculated, grayish-white, well-circumscribed soft tissue mass appeared inferior to the articular eminence. The peduncle of the soft tissue mass was firmly attached to the synovial membrane of the antero-lateral portion of the superior articular cavity (Fig. 4). The soft tissue mass, 18 × 15 × 10 mm in size, was removed.
with the attached synovial membrane (Fig. 5). The articular disc was conserved because its surface was macroscopically unaffected by the soft tissue mass.

3. Histopathological findings

The tumor, which was encapsulated by thin fibrous connective tissue, consisted of dense fibrous tissue including slit-like capillaries and ovoid or spindle-shaped cells without atypia or mitotic figures. Proliferated tumor cells were arranged in a focal storiform pattern. In addition, myxoid changes and multinuclear giant cells were not seen in the tumor (Fig. 6). Immunohistochemically, the ovoid or spindle-shaped tumor cell cytoplasm was immunoreactive to vimentin and alpha-smooth muscle actin, but not to S-100 protein, desmin or myosin (Fig. 7). Based on these histological and immunohistological findings, the intra-articular localized tumor was definitively diagnosed as FTS.

4. Postoperative Course

Postoperative healing was uneventful, and neither malocclusion nor facial nerve paralysis occurred postoperatively. TMJ function improved
gradually with time. Four weeks after surgery, left TMJ clicking and pain had resolved, and the maximum interincisal jaw opening increased to 52 mm. There have been no signs of recurrence in the 6 years since surgery.

**Discussion**

FTS was described initially by Geshickter and Copeland in 1949. In 1979, Chung and Enzinger reported detailed clinicopathologic findings of FTS in a study of 138 cases. The tumor could occur at any age, with a peak incidence at 20–50 years of age. Male patients outnumbered females with a ratio from 1.5:1 to 3:1. The tumor most frequently involves the fingers, hands, and wrists, and characteristically is a grayish-white, almost painless mass, is less than 20 mm in size, has a well-circumscribed, lobulated appearance, and attaches to the tendon sheath. Although a previous study provided proof of osseous changes, the tumor generally does not produce these changes. The tumor appears as a well-circumscribed mass on MRI, and the mass is seen as a low signal intensity lesion on T1-weighted images and as a low–high signal intensity lesion on T2-weighted images. Histologically, the tumor is encapsulated, and consists of a dense collagenous stroma containing slit-like capillaries and spindle-shaped fibroblast-like cells. Immunoreactivity for actin and the presence of actin-type microfilament bundles in the cytoplasm of these cells indicate that they are myofibroblasts, a consistent cellular component reported in FTS occurring in other parts of the body.

It has been reported that FTS most frequently arises from the tendon sheath or tendon, but rarely arises from other tissues. To our knowledge, five cases arising from the synovial membrane of the joint capsule have been reported in the English literature; two in the knee, one in the wrist, one in the radioulnar joint, and one in the TMJ. In the present case, the peduncle of the tumor was firmly attached to the synovial membrane of the TMJ capsule, and the tumor was localized within the TMJ. As for the origin of FTS, Pulitzer has suggested that FTS may represent a group of pathogenetically diverse lesions with similar clinical and histological features, and some of them may be derived from mesenchymal tissues other than tendon sheath.

In the present case, the tumor was finally diagnosed as FTS based on the imaging, macroscopic, histological, and immunohistological findings. Other benign tumors or tumor-like diseases arising from the synovial membrane of the TMJ, such as synovial hemangioma, synovial chondromatosis, giant cell tumor of tendon sheath, pigmented villonodular synovitis, and nodular fasciitis, have been previously reported. Because TMJ involvement with slow-growing tumors or tumor-like diseases, including FTS, could present with nonspecific signs and symptoms, attention should be paid to the imaging, macroscopic and histological features to distinguish FTS from these diseases. Synovial hemangioma involving the TMJ is rare, and its imaging features are unknown.
MR findings of synovial hemangioma arising in other parts of the body\textsuperscript{35} are similar to those of FTS. On arthroscopy, early-stage synovial hemangioma is recognized as a red or brownish-red mass\textsuperscript{36}. In contrast, FTS is recognized as a grayish-white mass. Therefore, arthroscopy may be effective for differentiating intra-articular localized FTS from synovial hemangioma. Several studies have reported the clinical and imaging features of synovial chondromatosis involving the TMJ. In synovial chondromatosis, undifferentiated cells of the synovial membrane transform into cartilage or bone, usually seen as multiple, small, calcified lesions on CT\textsuperscript{20, 21}, multiple-signal voids on T2-weighted MR images\textsuperscript{19}, and loose bodies in the articular cavity on arthroscopy\textsuperscript{20, 21}. Therefore, FTS is easily differentiated from synovial chondromatosis. Giant cell tumor of tendon sheath and pigmented villonodular synovitis are histologically very similar diseases, and it is rare for them to involve the TMJ\textsuperscript{22, 23}. These diseases may also contain some fibroblast-like or myofibroblast-like cells, but they are generally dominated by proliferating synovial cells, foamy histiocytes and multinuclear giant cells\textsuperscript{5, 9}. Therefore, these diseases are also easily distinguished histologically from FTS in most cases. However, Phalen\textsuperscript{27} graded giant cell tumor of tendon sheath based on the amount of stromal collagen fiber, and he reported that there were not many multinuclear giant cells in grade III, which has the highest amount of collagen fiber. Because this finding indicates that FTS may be the end stage of these diseases, it may be impossible to strictly distinguish FTS from these diseases\textsuperscript{28}. Histological distinction between cellular or myxoid variants of FTS and nodular fasciitis may also be difficult. However, nodular fasciitis appears suddenly, grows rapidly, and usually attains its full size in 3 to 6 weeks\textsuperscript{1, 11}. FTS, in contrast, is generally slow-growing. Therefore, lesions of nodular fasciitis are poorly circumscribed compared with the usual examples of FTS\textsuperscript{5}.

In the present case, the tumor was excised with the synovial membrane to which the tumor was attached. At the 6-year follow-up there were no signs of recurrence or complications. FTS has no aggressive potential, and there is no evidence that it can undergo malignant change. Thus any recurrence is most likely the result of incomplete excision of the lesion\textsuperscript{1, 9-11}.

When a benign tumor or tumor-like disease involves the TMJ, the most important clinical issue is to preoperatively distinguish these diseases from malignant tumors or cystic diseases, such as ganglion. Comprehensive diagnostic imaging based on CT, MRI, arthrography, and arthroscopy plays a very important role in identifying the nature of lesions involving the TMJ.

References