Synovial Hemangioma of the Temporomandibular Joint: Case Report

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Hemangiomas are benign vasoformative neoplasms or developmental conditions of endothelial origin. Synovial hemangiomas arise from a synovial lined surface within a joint space. This report describes a case of synovial hemangioma of the left temporomandibular joint (TMJ) in a 65-year-old male patient. Histologic examination confirmed the diagnosis of synovial hemangioma. This is a rare case series, as the only case of synovial hemangioma with TMJ localization previously described was reported in 1988. *J Oral Facial Pain Headache 2018;32:e45–e48. doi: 10.11607/ofph.1986*

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Synovial hemangiomas are uncommon lesions of soft tissue considered to be benign vascular tumors that primarily affect women in the second decade of life and most commonly involve the knee joint.[1] The only previously described case of synovial hemangioma involving the temporomandibular joint (TMJ) was reported by Atkinson et al in 1988.[1] The neoplasm may be completely symptom-free (casually discovered on a radiologic examination) or present signs and symptoms such as pain, limited mandibular kinetics, or pulsatile bleeding.[1] The presence of a phlebolith in the area is virtually diagnostic of hemangioma, and the aspiration of blood from the neoplasm supports the diagnosis of synovial hemangioma. This report describes a case of a 65-year-old male patient affected by synovial hemangioma of the left TMJ.

Case Report

A 65-year-old male patient, with a history of lower lobectomy of the left lung and adjuvant chemotherapy for primary lung adenocarcinoma 7 years before, was referred for evaluation of a 12-month history of progressively worsening pain in the left TMJ area that was intensified during chewing.

Neurologic and otologic diseases had been previously excluded.

An extraoral examination showed a swelling located in the left preauricular region with a soft consistency that was slightly painful to palpation. TMJ auscultation was negative and no muscle tenderness was present, but jaw movements were limited and the patient's maximum mouth opening (MMO) was 25 mm. An intraoral examination revealed a normal oral mucosa.

The patient received a total-body computed tomography (CT) scan, requested by an oncologist for the lung adenocarcinoma follow-up. The scan showed that the left intra-articular joint space was larger than the right one (Fig 1). The enhanced CT demonstrated slight uptake of dye by the mass, suggesting the possibility of a vascular lesion. No erosion or communication between the TMJ and temporal bone was observed. An intra-articular temporomandibular disorder (TMD) was immediately excluded because of the absence of a positive history for TMJ and because the clinical examination revealed no clicking, popping, and/or crepitation noise.

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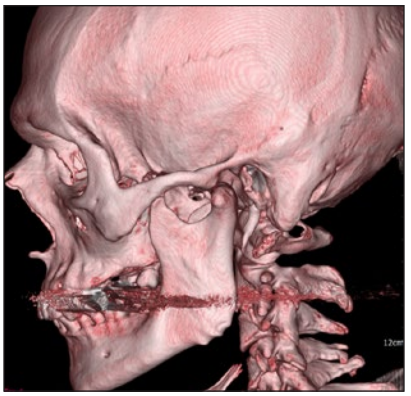


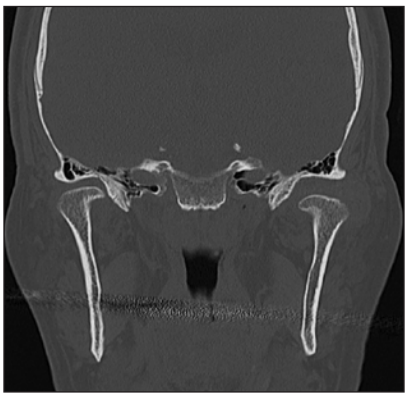
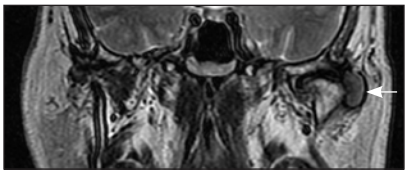


Fig 1 Computed tomography images.



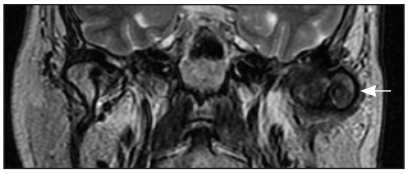


Fig 2 Magnetic resonance imaging, showing an irregular lesion at the left TMJ.

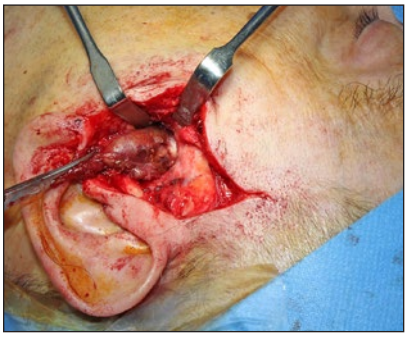


Fig 3 *(left)* En bloc excision of the mass.

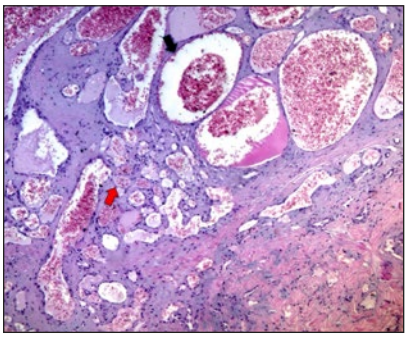


Fig 4 *(right)* Multiple large (black arrow) and small (red arrow) dilated vascular channels with thin walls lined by endothelium and filled with many red blood cells (hematoxylin and eosin staining, original magnification ×5).

Therefore, based on the Diagnostic Criteria for TMD (DC/TMD)[2] and the CT images, various types of TMJ lesions were suspected, including a malign neoplasm (such as a lung adenocarcinoma metastasis); a benign condition (such as chondromatosis, giant cell lesion, or synovial cyst); synovial hemangioma; or ganglion cyst.[3–7]

In order to improve the diagnostic work-up, magnetic resonance imaging (MRI) was requested to allow for a full assessment of the anatomical boundaries of the soft tissue lesion and of the mass intensity. The images showed an irregular lesion of 18 × 9 mm in the left condyle. The mass was characterized by a T1 hyperintense signal for vascular content (Fig 2) without abnormal calcifications. This suggested the possibilities of TMJ synovial chondromatosis or a giant cell tumor, as the MRI images did not show loose bodies or typical hypointensity on the T2-weighted images (T2WI).[1,8]

Exudation and accumulation of synovial fluid that can be observed on MRI and can help determine whether a lesion is of synovial origin were not present. The working hypothesis was that the mass was a synovial hemangioma.

Aspiration was not performed to minimize the risk of hemorrhage and to avoid mass reduction that would make surgical excision difficult. In November 2015, the patient was hospitalized for surgery. Under general anesthesia, a preauricular, pretragal skin incision was performed to approach the TMJ. After ligation of the superficial temporal vessels, further dissection revealed the mass to be confluent with the left TMJ capsule. En bloc excision of the mass was performed (Fig 3). The immediate postoperative period showed no complications, and the patient was dismissed 4 days later.

Histopathologic analysis revealed dense cellular connective and blood material. The connective tissue contained an inflammatory infiltrate characterized by lymphocytes (CD68+), differentiated lymphocytes (CD 45+), profuse hemosiderin pigment, and vascular structures (CD34+, CD31+). The proliferation index (Ki-67) was less than 1%. Immunohistochemistry was negative for thyroid transcription factor 1, cytokeratin 7, and cytokeratin 20. Based on these characteristics and in accordance with Bennett and Cobey,[9] the histopathologic diagnosis was of a localized intra-articular synovial hemangioma (Fig 4).

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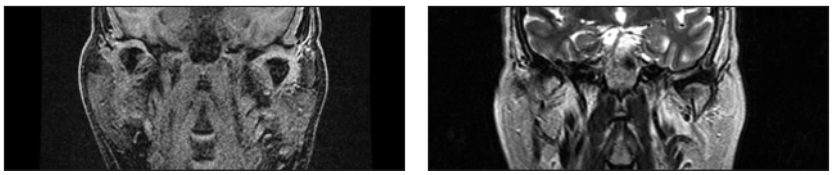

Fig 5 T1- and T2-weighted MRI images 12 months after surgery.

At 6 months postsurgery, the patient had normal masticatory function, was pain-free, and had an MMO of 35 mm. He was very satisfied with the treatment. No signs of recurrence were visible on the MRI images taken 12 months postsurgery (Fig 5).

Discussion

Synovial hemangiomas are uncommon, benign, vasoformative tumors of endothelial origin. TMJ synovial hemangiomas are very rare, with only one case report having been reported in the literature.[1] Therefore, synovial hemangioma classification is based on lesions in other joints.

According to Bennett and Cobey,[9] synovial hemangiomas can be classified into two general categories: localized and diffuse. The localized type is a well-circumscribed and pedunculated synovial mass, and excision of this type is quite simple. The diffuse type is infiltrative, with a clinical picture of intermittent joint pain, swelling, and hemarthrosis. The histologic features of the synovial hemangioma are cavernous lesions characterized by large, dilated, blood-filled vessels within an edematous, myxoid, hyalinized matrix.[10–12]

The differential diagnosis between high- and low-flow vascular lesions is crucial for successful treatment, as it decreases the occurrence of serious intraoperative hemorrhage.[6,10] Hemangiomas of the synovial membranes may also be classified by their juxta-articular, intra-articular, or mixed location. The intra-articular type, in which the tumor is lined by a synovial membrane, is the most common.

MRI is a useful tool for evaluating soft tissue lesions of the head and neck. It clearly shows the shape, size, signal intensity, and positional relationship of synovial hemangiomas to other tissues. An additional aid to diagnosis of synovial hemangiomas involves aspiration of blood from the tumor, producing a decrease in size.

Differential diagnosis should consider chondromatosis, giant cell lesions, and synovial and ganglion cysts. Synovial chondromatosis (SC) is a rare benign lesion with an intra-articular localization that can lead to the generation of a considerable number of osteochondromatous or cartilaginous nodes within the synovial fluid and intra-articular floating bodies.[13] It usually involves large joints, such as the knee; the TMJ is rarely affected (mostly the upper joint space). The tenosynovial giant cell tumor is a benign but locally aggressive tumor that arises from the synovial membranes of joints, tendon sheaths, and bursae, affecting predominantly the knee and hip. TMJ involvement has only been reported very rarely.[8] This tumor is characterized by deposition of hemosiderin, and MRI images are very helpful in its diagnosis because they show hypointensity throughout the lesion on T2WI. Synovial and ganglion cysts of the TMJ are uncommon; they are similar in clinical and radiologic presentation, but the anatomopathologic features are different. Synovial cysts are true cysts surrounded by cuboidal or flat synovial cells that may be connected with the joint cavity.[3] Ganglion cysts are pseudocysts encapsulated by fibrous tissue, without any connection to the joint cavity.[5]

Conclusions

The synovial hemangioma represents a rare lesion of the TMJ. However, it should be included in the differential diagnosis of TMJ neoplasms.

Acknowledgments

The authors report no conflicts of interest.

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