Radiology Case Reports 13 (2018) 499–502

Available online at www.sciencedirect.com

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journal homepage: http://Elsevier.com/locate/radcr

Recurrent pigmented villonodular synovitis of the temporomandibular joint

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ARTICLE INFO

Article history:
Received 2 October 2017
Received in revised form 22 November 2017
Accepted 1 December 2017
Available online 4 January 2018

Keywords:
Pigmented villonodular synovitis
Temporomandibular joint

ABSTRACT

Pigmented villonodular synovitis is a benign but locally aggressive extra-articular tumor arising from the synovial membrane of tendons and bursae occurring near a joint space. Rarely, pigmented villonodular synovitis can involve the temporomandibular joint, which is emphasized in this paper. Diffuse and localized types have been described in the literature. The diffuse type involves the entire synovial membrane and infiltrates adjacent structures, which tend to be more aggressive and associated with a higher rate of recurrence when compared with the localized type.

Case report

A 38-year-old woman initially presented to the otolaryngology clinic with complaint of a left-sided jaw mass. Contrast-enhanced computed tomography (CT) of the neck was notable for a large soft tissue mass involving the left masticator space partially invading and displacing the masseter, lateral pterygoid, and temporalis muscles. There was no evidence of erosion or destruction of the mandibular condyle or the left temporomandibular joint (TMJ) (Fig. 1). Further characterization with contrast-enhanced magnetic resonance imaging (MRI) was recommended, which showed a T1 iso- to hypointense mass in the left masticator space with heterogeneous enhancement measuring 7.5 × 6.4 × 5.5 cm in craniocaudal, transverse, and anteroposterior dimensions. There were multiple regions of T2 hypointensity within the central aspect of the mass (Figs. 2 and 3). The mass effect on the parapharyngeal space demonstrated medial displacement with infiltration and enhancement of all the muscles of mastication. High T2 signal was noted in the mandible but without cortical destruction or evidence of invasion. Additionally, there was suspicion for involvement of the trigeminal nerve with expansion of the foramen ovale. After biopsy and surgical excision of the soft tissue mass, pathology demonstrated large nodules of solid sheets of tumor cells with hemosiderin pigment admixed with xanthoma cells and a few multinucleated giant cells (Figs. 4 and 5). Immunohistochemical staining with CD68 was diffusely positive, confirming histiocyte origin (Fig. 6). No cellular atypia was reported. Findings were consistent with pigmented villonodular synovitis (PVNS), also known as giant cell tumor of the tendon sheath-diffuse type.

The patient was then referred to our institution 8 years later with complaint of left neck swelling and jaw pain for about 4

Competing Interests: The authors have declared that no competing interests exist.
Pathology images provided by Dr Reina Tarabey, MD, Mercy Hospital Pathology Department, Chicago, IL.

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https://doi.org/10.1016/j.radcr.2017.12.001

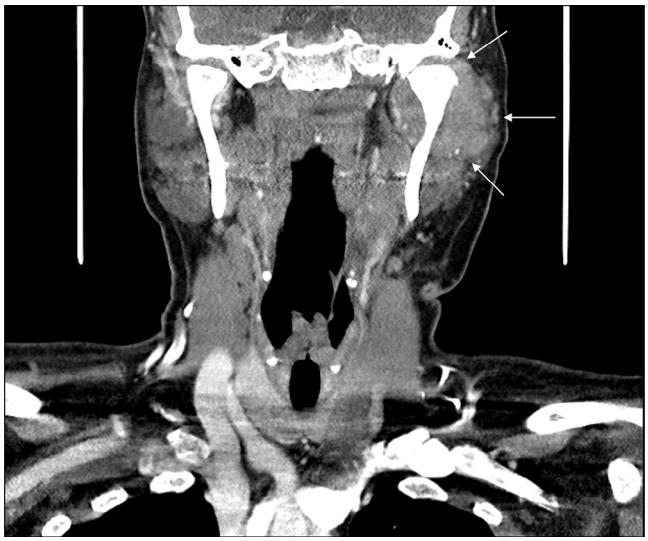

Fig. 1 – Contrast-enhanced coronal computed tomography shows a solid enhancing mass in the left masticator space surrounding the mandible and displacing the parapharyngeal space.

months. Physical exam demonstrated a hard, firm, mobile 3-cm mass near the left parotid gland. Repeat contrast-enhanced MRI demonstrated a heterogenous mass with central hypointensity on T1- and T2-weighted images involving the left masticator space and wrapping around the mandibular ramus measuring 2.8 × 4.4 × 4.2 cm in anteroposterior, transverse, and craniocaudal dimensions without appreciable enhancement (Figs. 7-9). There was minimal signal change in the mandible itself without enhancement. A fine needle aspiration was then obtained showing multinucleated giant cells with hemosiderin-type pigment and stromal cells highly suggestive of recurrent PVNS. A complete resection with TMJ reconstruction and anterolateral thigh free flap reconstruction is currently preoperatively planned.

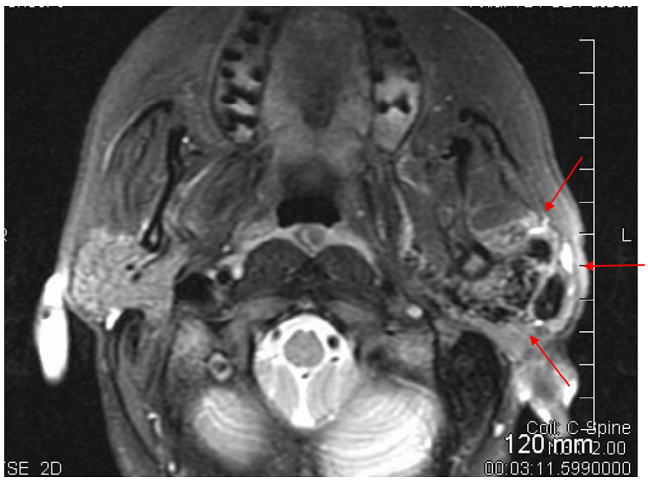

Fig. 2 – Axial T2-weighted image shows multifocal regions of low signal intensity within the central mass involving the left masticator space and temporomandibular joint.

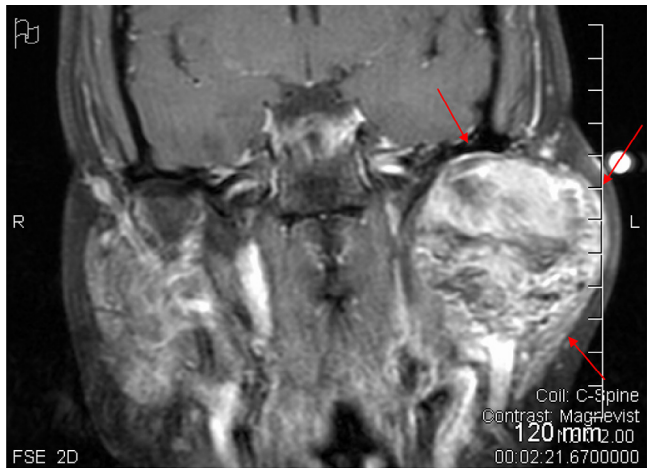

Fig. 3 – Coronal postcontrast T1-weighted fat-saturated image shows a solid mass in the left masticator space with areas of low signal intensity and mild heterogeneous enhancement.

Discussion

PVNS involving the TMJ is a rare entity and is difficult to diagnose clinically. The reported incidence is 1.8 annual cases per million individuals in which most lesions affect the knee, hip, or shoulder [1]. Patients usually present in the third and fourth decades with symptoms of TMJ dysfunction [2]. Imaging, particularly utilizing MRI, is helpful as it can demonstrate a characteristic hemosiderin pattern and outlines the extent of the mass to aid in treatment and prognosis. When the TMJ is involved, most cases are of the diffuse histologic type. The etiology of this disease process is unclear, with histopathology characterized by hyperplasia of the synovium in joints and tendinous sheaths. There is accentuated proliferation of the stroma cells, and a significant quantity of intracellular and extracellular

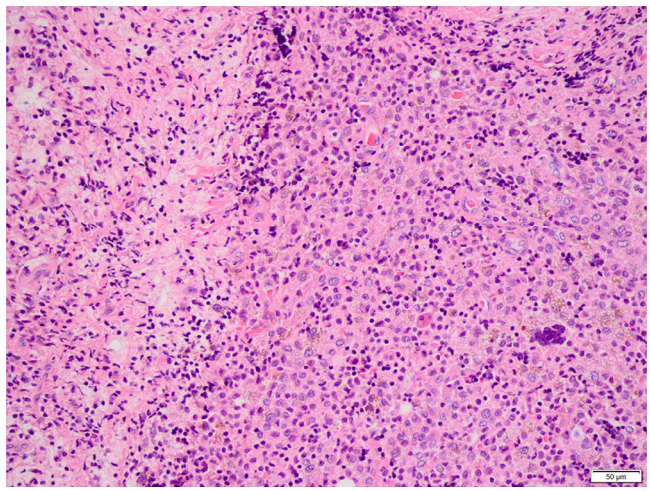

Fig. 4 – Hematoxylin and eosin of the tumor demonstrating solid sheets of cells separated by fibrous stroma. Scattered histiocytes and multinucleated giant cells contain intracytoplasmic hemosiderin.