B-ENT, 2015, **11**, 287-289

Inflammatory pseudotumour of the temporomandibular joint

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Key-words. Inflammatory pseudotumor; temporomandibular joint

Abstract. *Inflammatory pseudotumour of the temporomandibular joint*. Head and neck inflammatory pseudotumors (IPs) are rare, idiopathic, non-neoplastic lesions that most commonly affect the orbit, but may involve other areas such as the larynx, oropharynx, paranasal sinuses, and meninges. We report the case of a 55-year-old man who presented with progressive left-sided hearing loss, aural fullness, and otalgia. Computed tomography and magnetic resonance imaging (MRI) detected a soft-tissue mass in the left temporomandibular joint (TMJ). Histopathologic examination showed overlying squamous epithelium with hyperkeratosis, parakeratosis, subepithelial fibrosis, and chronic inflammatory infiltrate, which were consistent with an IP. Radiologic images and MRI indicated an ill-defined soft tissue involving the roof and posterior aspect of the TMJ, extending into the anterior external auditory canal. Our case was treated with a 2-week course of high dose prednisone (1 mg/kg) and a 2-week taper with resolution of symptoms. Two years after treatment, the patient shows no evidence of recurrence on MRI.

Introduction

Pseudotumors are most commonly found in the lung, thorax, and gastrointestinal tract, but have been described in almost any location.[1,2] Head and neck inflammatory pseudotumors (IPs) are rare, and most commonly affect the orbit, representing 6-9% of all orbital tumors.[3] However, they may involve other areas such as the larynx, oropharynx, paranasal sinuses, and meninges.[2,4] It is a diagnostic challenge to differentiate these lesions from neoplasms, and the diagnosis is often made by exclusion of other lesions.[3]

Case presentation

A 55-year-old man presented with increasing left-sided hearing loss that lasted 7 weeks, aural fullness, and otalgia. He had no other medical problems, with no otologic infections or surgery and no family history of hearing disorders. Physical examination revealed an intact tympanic membrane (TM), with a soft tissue lesion in the anteromedial aspect of the ear canal abutting the TM. The patient had radiating pain in the jaw, muscle stiffness, limited movement of the jaw, and clicking of the temporomandibular joint (TMJ) when opening the mouth. These symptoms persisted after surgery. The mass appeared to move with movement of the mandible. Audiometric testing confirmed left-sided conductive loss.

A temporal bone computed tomography (CT) scan revealed a soft tissue nodule at the anterior and inferior aspects of the external auditory canal (EAC) abutting the TM, measuring 0.4 cm anterior-posterior × 0.6 cm transversal × 0.4 cm craniocaudal (Figure 1). A post-gadolinium axial T1-weighted magnetic resonance image (MRI) of the left internal auditory canal demonstrated a 1.5 × 1 cm lesion in the area of the EAC and TMJ with focal erosion into the floor of the bony EAC (Figure 2). A transaural incisional biopsy of the lesion was performed and histology showed overlying squamous epithelium with hyperkeratosis, parakeratosis, subepithelial fibrosis, and chronic inflammatory infiltrate, suggesting an IP. This case was not reviewed by our institutional review board because it considers single case reports exempt.

Discussion

In two literature reviews of space occupying lesions of the TMJ from 1933-1999 (including over 285 TMJ tumors), there were no cases of IP identified.[2,3]

Figure 1
Axial CT of temporal bones at the level of the ear canal on the left side. The image shows a TMJ-based mass protruding into the medial ear canal (arrow).

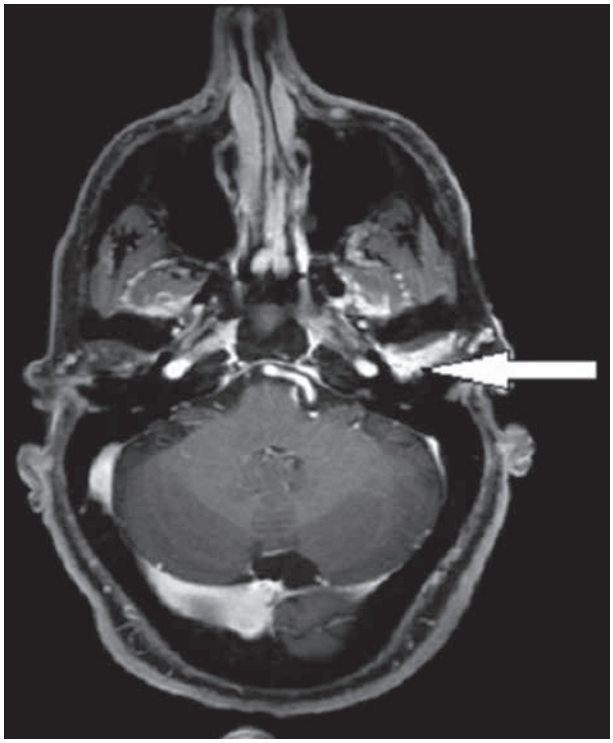

Figure 2
Axial T1-weighted post-gadolinium MRI showing a hyperintense mass in the TMJ extending into the medial ear canal (arrow).

A current review of English peer-reviewed literature shows no TMJ localized cases of IP. There are two cases of IP that presented with TMJ dysfunction; however, the IP was from an orbital source to the infratemporal fossa[4] in one case and in the second case the IP was extensive involving the masticator space, the pterygoid and temporalis muscles, the maxillary sinus, and the infratemporal fossa.[5]

Common TMJ disorders usually present with either pain, disturbed or restricted opening of the mouth, and joint sounds. Rarely, TMJ lesions may also present with conductive hearing loss, dental malocclusion, or pain that is not associated with mandibular movements.[4,5] Prompt diagnoses of TMJ tumors or pseudotumors may be difficult as they present similarly to patients with TMJ dysfunction. Histologically, these IP lesions appear as inflammatory infiltrate with a fibrotic background. Clinical presentations vary from slow growth with minimal mass effect to bony destruction that can mimic malignancy. However, 71.6% of space occupying lesions of the TMJ are non-inflammatory pseudotumors, including pathologies such as osteochondromas, synovial chondromatosis, pigmented villonodular synovitis, and eosinophilic granulomas; while, 18% of TMJ lesions are malignant tumors, such as sarcomas and metastatic disease.[3]

The radiologic findings of IPs may be nonspecific. On CT, a moderately enhancing mass is usually seen, with possible soft tissue or bony erosion, remodeling, and sclerosis.[3] On MRI, the lesion is usually hypointense to isointense compared with muscle on T1-weighted images and is usually relatively hypointense on T2-weighted images. The hypointensity may be due to the relative lack of mobile protons within fibrotic lesions. Often, there is moderate to marked post gadolinium injection enhancement.[1,2] The enhancement usually has a delayed pattern, which may be due to the accumulation of extravascular contrast medium in fibrotic lesions.[1]

Total or subtotal surgical resection is preferred for operable cases and in cases with bony destruction.[2] The most common surgical complications in TMJ surgery are mandibular deviation,

decreased range of motion, and facial palsy.[3,4] High-dose steroid or radiation therapy is often reserved for inoperable tumors, such as those involving the orbit.[3,5,6] More information is available for orbital pseudotumors, as high-dose corticosteroids (prednisone 1 mg/kg or equivalent for 2 weeks, followed by taper) are successful in controlling >50% of orbital diseases.[3,5]

This patient was treated with a 2-week course of high dose prednisone (1 mg/kg) and a 2-week taper with the resolution of symptoms. Two years after treatment, the patient has no evidence of recurrence on MRI. Aural fullness and hearing loss have improved; although, herniation of TMJ contents into the EAC continues due to the partial loss of the TMJ posterior wall. High-dose steroids may be the preferred method of treatment for small TMJ IPs, with surgery or radiation reserved for non-responsive lesions.

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