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Pigmented Villonodular Synovitis of the Temporomandibular Joint: A Radiologic Diagnosis and Case Report

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Pigmented villonodular synovitis (PVNS) is a benign, but locally aggressive, proliferative disorder arising from the synovium, often with infiltration and/or osteoinvasion. It commonly arises in the tendon sheaths and long bone joints and, rarely, in the bursae. It is usually a monoarticular disease, with involvement of the knee joint in 80% of cases and then, in decreasing order of frequency, the hip, ankle, small joints of the hands and feet, wrist, shoulder, and elbow.[1-3] PVNS of the temporomandibular joint (TMJ) is rare, with approximately 46 cases reported to date.

The earliest published reference to articular PVNS was by Chassaignac in 1852.[4] He described it as a nodular lesion of the flexor digital tendons (known then as a giant cell tumor of the tendon sheath). In 1941, Jaffe et al[5] introduced the currently accepted histopathologic features and established classification of the lesions under the umbrella term "pigmented villonodular synovitis."[6,7] They divided PVNS into the more common diffuse form (affects the entire synovial joint membrane or bursa) and the localized form (affects only a portion of the joint lining or tendon sheath with a solitary pedunculated, nodular lesion with no villi).[8-10] The first report of TMJ PVNS was by Lapayowker et al[6] in 1973.

The present report describes a case of unilateral TMJ PVNS and the role of radiologic imaging, in particular, magnetic resonance imaging (MRI), in establishing the diagnosis.

Case Report

A 51-year-old woman was referred with a 20-year history of left otitis externa and a 2-month history of left jaw and neck pain associated with progressive preauricular swelling. She reported intermittent restriction of mouth opening that improved when taking antibiotics. She had no associated symptoms of infection, and previous treatment with oral ciprofloxacin and corticosteroid/antibacterial ear drops had only partially relieved her symptoms. She had a history of depression and breast cancer with no metastatic disease.

The initial examination revealed a swollen left preauricular region and tenderness within the left TMJ, temporalis muscle and coronoid process. She had full mandibular range of motion but the follow-up examination revealed progressively limited mouth opening. Her facial nerve function was not affected.

Her erythrocyte sedimentation rate was mildly elevated at 29 mm/hr. Autoimmune markers, including rheumatoid factor, were negative. A contrast-enhanced computed tomography scan demonstrated thickened and enhancing tissue around the left mandibular condyle (Fig 1). The adjacent lateral pterygoid muscle was enlarged (Fig 1). Assessment of the bone windows revealed a flattened and sclerotic left condyle with large rounded cystic erosions. The glenoid fossa was also abnormally sclerotic (Figs 2, 3). The findings were consistent with inflammatory/erosive arthropathy of the TMJ, with osteomyelitis and TMJ septic arthritis the most likely etiology. MRI was obtained to further define the abnormality, and positron emission tomography requested because of her history of breast cancer. The MRI scan showed significant left TMJ arthropathy with prominent synovitis and a small effusion (Fig 4). The thickened synovium demonstrated T_1- and T_2-weighted hypointensity (Figs 5, 6). Assessment of the planning sequence, performed with a gradient echo technique, demonstrated susceptibility artifact around the left TMJ, suggesting the presence of blood products (Fig 7). A decreased marrow signal in the mandibular condyle and glenoid fossa was consistent with sclerosis (Fig 6), and small osseous erosions were present

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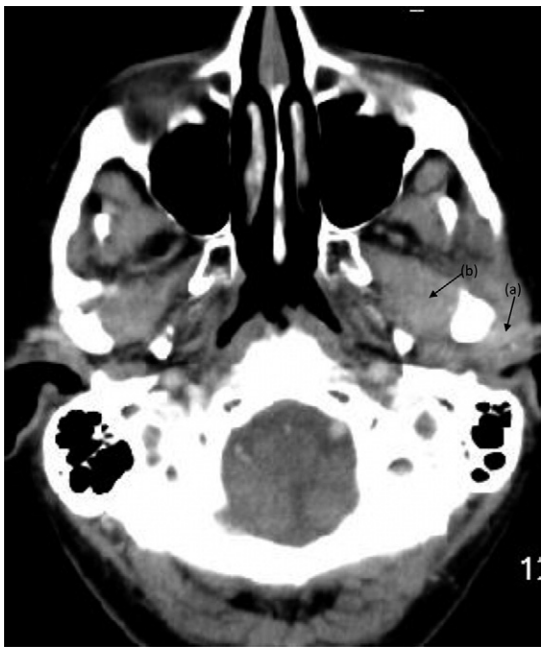

FIGURE 1. Contrast-enhanced computed tomography image demonstrating prominent enhancing synovium around left mandibular condyle (*a*) and enlargement of adjacent lateral pterygoid muscle (*b*).

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(Fig 8). No parotid, temporal bone, or intracranial pathologic features were noted. Overall, the findings were consistent with inflammatory arthropathy of the left TMJ, with the presence of prominent, likely hemorrhagic synovitis, highly suggestive of PVNS. The positron emission tomography scan revealed low to moderate grade joint-based tracer uptake involving the left TMJ. No evidence was found of metastatic disease. The uptake in the left TMJ was consistent with an arthritic etiology.

The patient underwent open exploration of the left TMJ with incisional biopsy. An extended preauricular approach[11] was used to expose the left TMJ. The mass was intracapsular with capsular expansion. The capsule was opened, and a gelatinous, soft, red-brown tissue filled the joint space. An incisional biopsy was sent for histopathologic and microbiologic examination. The specimen showed fibrofatty tissue containing mildly cellular areas with prominent hemosiderin deposits and scattered bland giant cells (Fig 9). No evidence of infection or malignancy was found. No growth occurred on culture. A histologic diagnosis of PVNS was confirmed.

Definitive management involved thorough resection of the left TMJ PVNS using the previous approach. Total synovectomy, discectomy, and curettage of the condylar head and glenoid fossa were performed. Careful inspection revealed no communication with the middle cranial fossa. An interpositional temporalis turndown flap was fashioned over the zygomatic arch, and the incision was closed in layers. Histopathologic examination of the definitive specimen confirmed the diagnosis of PVNS, revealing a hyperplastic synovium with areas of fibrosis and subsynovial proliferation of bland mononuclear cells with oval nuclei and focal hemosiderin. Rare multinucleated giant cells were present (Fig 9).

The postoperative recovery was uneventful, except for a mild increase in pain and swelling over the left TMJ on maximal mouth opening at 2 months. This resolved with amoxicillin and anti-inflammatories. At 1 year postoperatively, she had no residual pain, no facial nerve dysfunction, normal occlusion, and painless TMJ range of motion with a maximal interincisal opening of 38 mm and lateral excursion of 9 mm bilaterally.

Discussion

TMJ PVNS is a rare disorder with an annual incidence of 1.8 per million population.[9] It occurs in all age groups but more commonly in the third to fifth decades.[8] No gender predilection exists. There are many hypotheses of the etiology of PVNS. The most widely accepted is a chronic, reactive inflammatory response of the synovium of unknown origin, in which the histiocyte plays a pivotal role. Other theories include altered lipid metabolism, benign neoplastic tissue proliferation of synovial, vascular, or fibrohistiocytic origin, and trauma resulting in repetitive intra-articular hemorrhage that eventually manifests as hemosiderin deposition. Our patient had a chronic history of presumed ipsilateral otitis externa that might have masked an earlier diagnosis of TMJ PVNS.

The symptoms can include altered hearing, otalgia, a progressive preauricular mass or swelling, often minimal pain, malocclusion, paresthesia, and temporomandibular symptoms, including clicking and limited mandibular range of motion. Otalgia, preauricular swelling, neck pain, and progressive trismus were experienced by our patient.

Of the reported cases, 71% involve bony erosion,[12] in particular with the diffuse form of PVNS.[3,10] Our case exemplified a diffuse form of TMJ PVNS with extensive erosive arthropathy. The

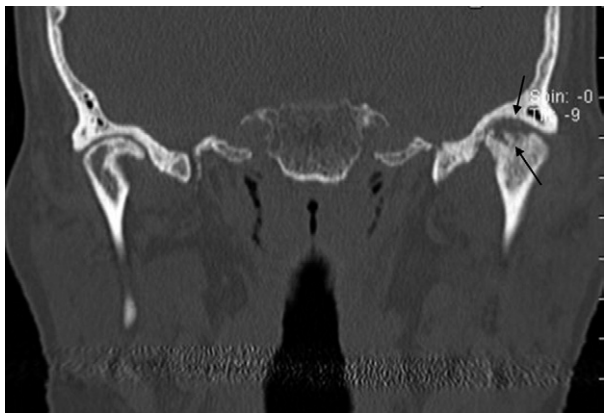

FIGURE 2. Coronal computed tomography image on bone window demonstrating sclerosis of left mandibular condyle and glenoid fossa with focal erosions in mandibular condyle (*arrows*).

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