Abnormality of the mandible ramus and a tumor in the parotid region

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CLINICAL PRESENTATION

In January 2011, a 36-year-old Chinese man was admitted to the Department of Oral and Maxillofacial Surgery, Affiliated Stomatology Hospital of Kunming Medical University, with a 6-month history of a progressive abnormality in the left cheek and restriction of mouth opening to no more than 1.0 cm (Figure 1). On admission, the patient's body temperature was 36.4°C; pulse, 72 beats per minute; respiration rate, 18 breaths per minute; and blood pressure, 110/85 mm Hg. His medical history indicated that he had not had temporomandibular joint (TMJ) problems or any limitations of opening by antecedent trauma or tooth extraction. Upon physical examination, a solid mass was found in the left parotid gland; no facial paralysis and no crepitus or clicking were found in either TMJ. In addition, lymph nodes were not enlarged in the cervical and supraclavicular areas. Routine laboratory analysis was as follows: white blood cell count, 7.62×10^9/L; hemoglobin level, 175 g/L; red blood cell count, 5.93×10^{12}/L; platelet count, 242×10^9/L; prothrombin time, 12.2 seconds; international normalized ratio, 1.29; fibrinogen, 2.96 g/L; thrombin time, 16.5 seconds; and activated partial thromboplastin time, 45.0 seconds. A panoramic radiograph found that the condyle was intact and smooth except for the resorption of the mandibular ramus. A computed tomography (CT) scan of the jaw showed that the articular space was normal but that there were 3 lesions on the left side: 2 soft tissue lesions and atrophy of the mandible. The first soft tissue lesion was located in the parotid gland and was associated with a high-density area. The second soft tissue lesion was located in the left lateral and inferior condylar process area, with a low-density area in the central region. In addition, there were 2 dome-shaped masses located in the maxillary sinuses, which were consistent with chronic sinus inflammation (Figures 2 to 4).

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DIFFERENTIAL DIAGNOSIS

The clinical and radiographic presentation suggested 3 separate and distinct disease processes. The differential diagnosis for the parotid lesion included benign salivary neoplasms (such as pleomorphic adenoma), low-grade malignant salivary neoplasms (such as adenoid cystic carcinoma and mucoepidermoid carcinoma), and squamous cell carcinoma. The differential diagnosis for the lesion associated with the TMJ included pigmented villonodular synovitis (PVNS) and synovial chondromatosis. The abnormality in the left cheek and mandible ramus was considered to be an independent disease, different from the parotid gland lesion. The ramus lesion was thought to be progressive facial hemiatrophy.

Because the physical examination and CT scan found that the solid mass was located in the left parotid gland, with well-circumscribed margins, salivary pleomorphic adenoma was high on the differential diagnosis. Pleomorphic adenomas (benign mixed tumors)[1-3] primarily arise in the major salivary glands, especially in the parotid, mostly with no accompanying facial nerve weakness and no restriction of mouth opening. Radiologically, the lesion presents as a unilobar or multilobar high-density area, with clear demarcation. However, in this case, the complaint of the abnormality in the left cheek and the restriction of mouth opening to no more than 1.0 cm made this diagnosis less likely. There were other possible diagnoses, such as mucoepidermoid carcinoma,[4] adenoid cystic carcinoma,[5] and squamous cell carcinoma,[6] which develop primarily in the deep portion of the parotid gland and then spread to extraparotid areas such as the TMJ, leading to a complaint of restricted mouth opening. However, the radiologic examination showed the lesion with a distinct margin, which is not consistent with the common manifestations of malignant tumors in the parotid area.

In assessing the lesion adjacent to the TMJ, consideration was given to the differential diagnosis of TMJ lesions, such as PVNS and synovial chondromatosis. PVNS,[7,8] which is an uncommon, benign, locally invasive lesion, arises from the synovial membranes of particular body joints, bursae, and associated tendon sheaths and most commonly occurs in the hip, ankle, small joints of the hands and feet, shoulder, and elbow. In fact, any synovial-lined articulation may be affected.

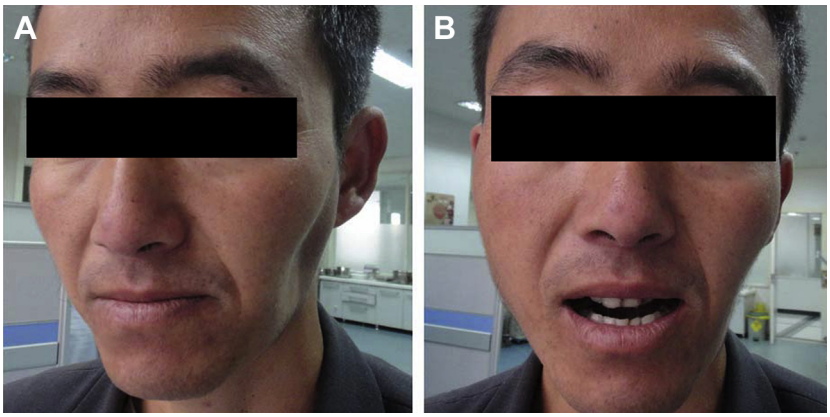

Fig. 1. **A**, **B**, Facial asymmetry and restriction of mouth opening to no more than 1.0 cm.

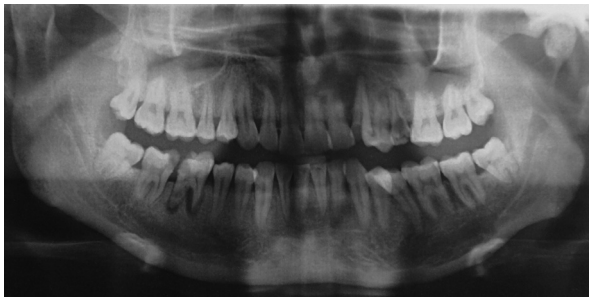

Fig. 2. Panoramic radiograph showed extensive resorption of the left mandibular ramus, as well as soft tissue proliferations of the maxillary sinuses.

If PVNS occurs in the TMJ, the majority of symptoms related to the TMJ or TMJ dysfunction include a local sensation of heat and tenderness, trismus, and TMJ clicking. In addition, its clinical presentation can mimic a parotid tumor, with a painful or painless preauricular mass. PVNS is related to a family of lesions including pigmented villonodular tenosynovitis and pigmented villonodular bursitis, with the diagnosis depending on a radiologic and pathologic examination. Normally, with PVNS, a CT scan shows a normal masticator space and a well-marginated tumor, but with bone erosion.[9] A magnetic resonance imaging scan may find low signal intensity (T2-weighting) and a "blooming" artifact from hemosiderin within the lesion (gradient-echo sequences).[8] In the present case, the lesion in the left lateral and inferior condylar process favored the characteristics of PVNS. However, no crepitus or clicking was found in either TMJ. In addition, a CT scan of the jaw found that the condyle was intact and smooth. These considerations argued against a diagnosis of PVNS.

Synovial chondromatosis of the TMJ, which is considered to be a benign lesion and is characterized by metaplasia of the connective tissue leading to chondrogenesis in the synovial membrane, presents as multiple hyaline cartilage nodules typically within subsynovial tissues. Although the etiology is not well known, repetitive trauma, rheumatoid arthritis, and an inflammatory or noninflammatory arthropathy may not be excluded. On rare occasions, synovial chondromatosis of the TMJ presents a preauricular swelling similar to a sign of a parotid tumor and may be mistakenly diagnosed and treated as a parotid mass.[3,10] A CT scan will normally show multiple irregularly shaped calcifications.[11] Although in this case the lesions in the left lateral condyle involved the TMJ, the possibility of synovial chondromatosis was not favored because no focal ossification was presented, but instead a low-density area was discovered in the central portion of the lesion. Therefore, this diagnosis also was excluded.

Owing to the abnormality in the left cheek and the mandible ramus, Parry-Romberg syndrome (PRS) was suspected as an independent disease from the parotid gland lesion. PRS, also known as progressive hemifacial atrophy, is an uncommon disorder characterized by a slowly acquired progressive and unilateral facial atrophy involving skin, subcutaneous tissue, muscles, cartilage, and bony structures.[12] Most of the time, progressive shrinking and deformation of one side of the face results in unilateral facial atrophy, as well as deviation of the mouth and nose toward the affected side. In addition, mandibular involvement is common in PRS. A CT scan, along with the mirror image of the unaffected side, is very helpful in assessing the degree of asymmetry.[12,13] In our case, the abnormality in the left cheek and the mandible ramus suggests a diagnosis of PRS. However, it is important to note that the lesion in the parotid region was not adjacent to the abnormality in the cheek and the mandible ramus.

In summary, the clinical features, radiographic findings, and medical history of the patient were most consistent with a low-grade malignant tumor of the parotid, which extended to the TMJ area, resulting in the restricted mouth opening.