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CASE REPORT

Mucinous adenocarcinoma of the temporal region initially diagnosed as temporomandibular disorders: a case report

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Adenocarcinoma occurring in the temporal region has not previously been reported. We present a case of mucinous adenocarcinoma of the temporal region. A 62-year-old female patient was diagnosed as having temporomandibular disorders because of severe trismus and joint pain. Although trismus progressively worsened, there were no abnormal findings on diagnostic imaging studies including magnetic resonance imaging (MRI) and bone scintigraphy. As swelling of the temporal region was observed, biopsy was performed. Histologic examination showed chronic inflammation of the striated muscle. Approximately 6 months later, follow-up MRI demonstrated an ill-defined mass lesion in the infratemporal region extending to the intracranium. Histologic diagnosis of the biopsy showed that this mass lesion was moderately differentiated mucinous adenocarcinoma.
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Keywords: mucious adenocarcinoma; temporal region; temporomandibular disorders

A 62-year-old female patient was referred to our department in October 2003 with a history of right facial pain and severe trismus. Eight months earlier she had been referred to another clinic for evaluation of trismus and temporomandibular joint (TMJ) pain, which had been present for 2 years. She was diagnosed as having temporomandibular disorders (TMD) and underwent occlusal splint therapy for about 6 months. However, trismus progressively worsened and the joint pain persisted. At the initial consultation in our department, she complained of pain in the right temporal region as well as in the pre-auricular area during mouth opening and chewing. Mild tenderness was present in the right temporal muscle, but a mass was not palpable. In addition, neurogenic facial pain or migrainous headache was not observed. Maximal mouth opening was 13 mm, and passive stretching was ineffective. A panoramic radiography did not show any change in the right mandibular condyle or mandibular fossa. Lateral oblique transcranial projections showed no hinge movement. On magnetic resonance imaging (MRI), the right TMJ disc appeared slightly compressed, but the disc position in relation to the condyle was normal (Fig. 1). Laboratory examinations revealed that LDH was slightly elevated, but the white blood cell count and C-reactive protein (CRP) were within the normal range. There were no evidences of the parotid gland enlargement as demonstrated by MRI and no abnormal findings on salivary secretion. A tentative diagnosis of the internal derangement of TMJ attributed to a 'stuck disc' was made. However, arthrocentesis to improve the limitation of condylar movement and relieve the pain was ineffective.

In December 2003, the patient noticed a swelling in the right temporal region and slight paresthesia extending from the right lower eyelid to the upper lip. The Gd-DTPA enhanced T1-weighed MRI images showed irregular enhancement of the temporal muscle in the infratemporal fossa and the adjacent meninges (Fig. 2). Bone scintigraphy with Tc-99 m revealed increased uptake extending from the right temporal bone to the maxilla (Fig. 3). Based on these findings, either temporal myositis or osteomyelitis of the temporal bone were suspected, but the etiology remained uncertain. The patient was maintained on antibiotic therapy for 2 weeks with no apparent improvement of the symptoms. Therefore in February 2004, a biopsy of the right temporal muscle was performed. Microscopic examination showed chronic inflammation of the striated muscle, with prominent round cell infiltration, and especially predominant lymphocyte accumulation surrounding the capillary vessels. Immunoassay for complement component-1 (C1) and complement component-4 (C4) detected increases in these components in the serum, indicating the possibility that the lesion represented an immune complex disease such as a vasculitis. Accordingly, a

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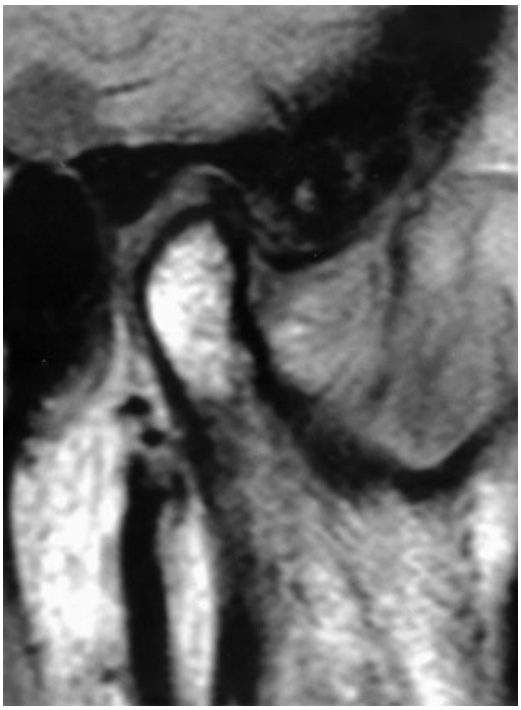

Figure 1 Sagittal MR image of the right TMJ at the initial consultation in October 2003. Although the disc appears to be slightly compressed, the position of the disc in relation to the condyle is normal.

steroid was administered for 1 month, but this regimen was only minimally effective in reducing the swelling.

In April 2004, follow-up MRI demonstrated an ill-defined mass lesion measuring 2.5 × 1.0 cm in the right infratemporal region extending to the intracranium (Fig. 4). An extraoral biopsy was carried out and showed the lesion to be a moderately differentiated mucinous adenocarcinoma (Fig. 5). Ga-67 scintigraphy showed metastasis to the vertebra. Because of the extent of the process, radiation therapy and chemotherapy were planned as palliative treatment. However, the patient and her family declined further treatment, and she died 6 months later.

Discussion

Patients who are referred to a clinic with a major complaint of limited mouth opening and pain in the pre-auricular region are often diagnosed as having TMD, and undergo TMD therapies. However, if not responsive to these therapies, the possibility of diseases other than TMD cannot be ruled out. Neuromuscular diseases must be considered in the differential diagnosis of trismus. However, when swelling in the right temporal region and infraorbital paresthesia developed in this case, it was suggested that the trismus did not originate from such neuromuscular diseases on the scintigraphic findings. It has been reported that tumors occurring in the TMJ or pre-auricular region demonstrated TMD-like manifestations in the development of pain and trismus and such TMD-like symptoms may represent insidious malignant processes (1, 2). Therefore, although the initial symptoms in the present case were quite similar to those of TMD, definitive diagnosis should be established by paying careful attention to other clinical signs that are not observed in TMD.

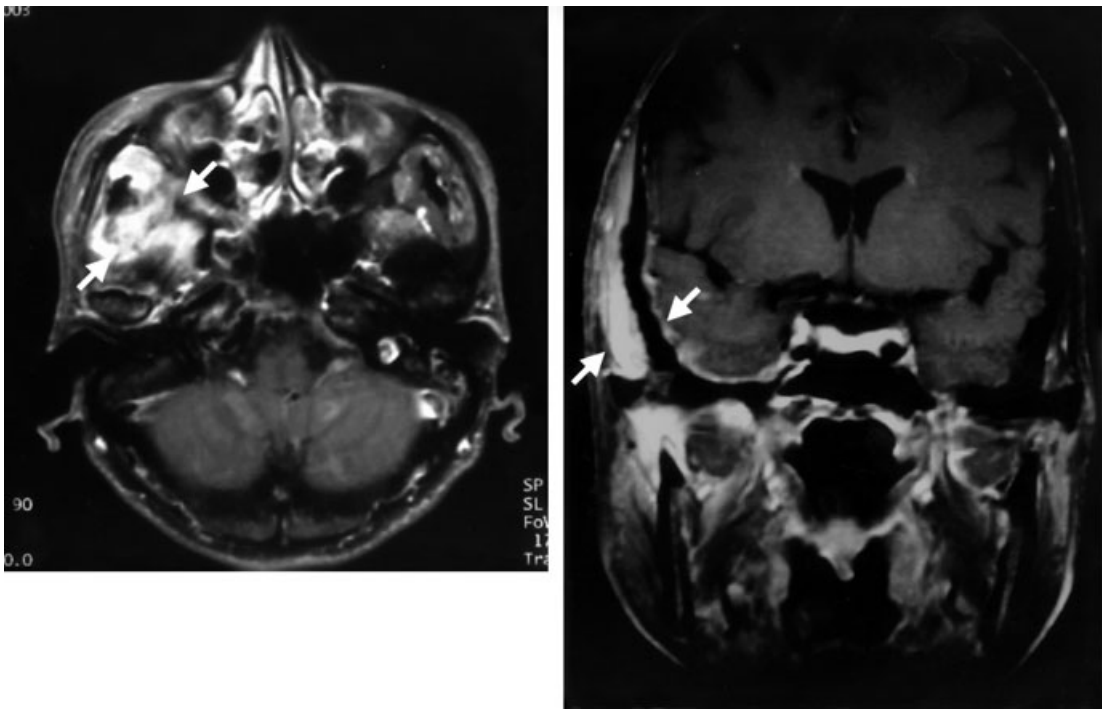

Figure 2 T1-weighted MR images enhanced with Gd-DTPA in December 2003. Horizontal plane (left) and the coronal plane (right). The arrows indicate areas in the infratemporal fossa and the adjacent meninges of that are enhanced.

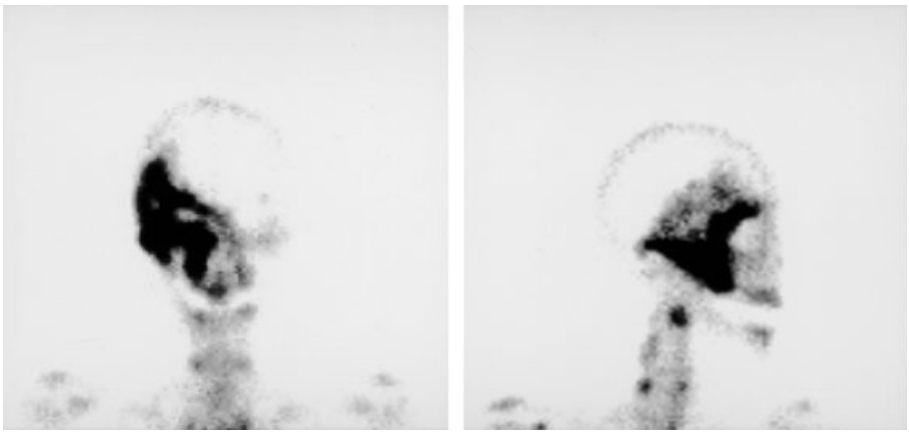

Figure 3 Bone scintigraphy with Tc-99 m showing marked uptake in the area from the right temporal bone to the maxilla.

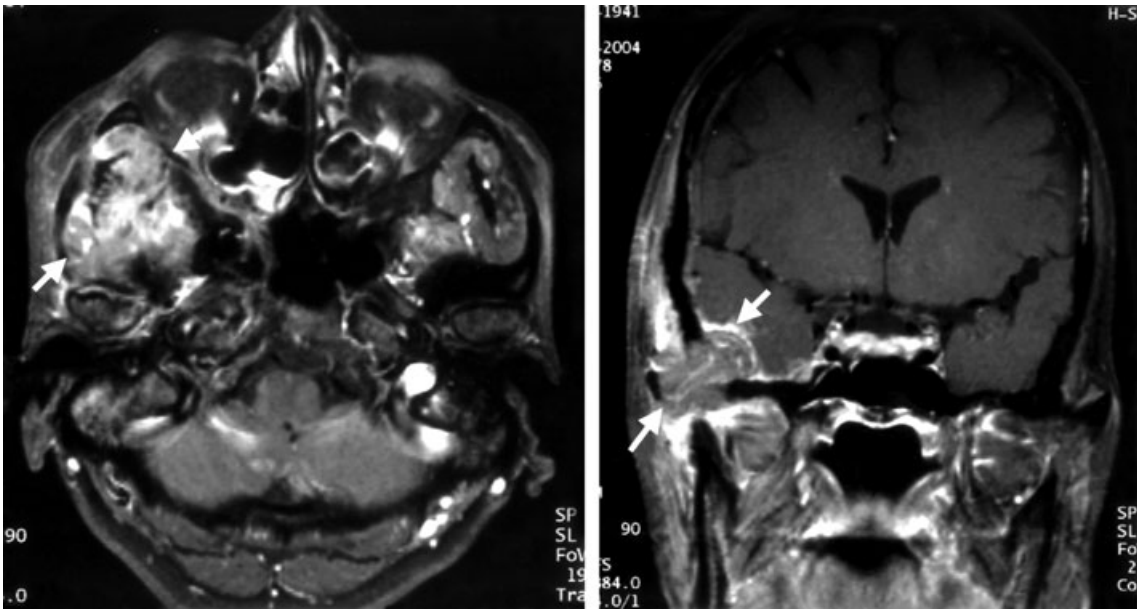

Figure 4 T1-weighted MR images enhanced with Gd-DTPA in April 2004. Horizontal plane (left) and coronal plane (right). The arrows indicate a mass lesion with ill-defined margins in the right infratemporal region extending to the intracranium.

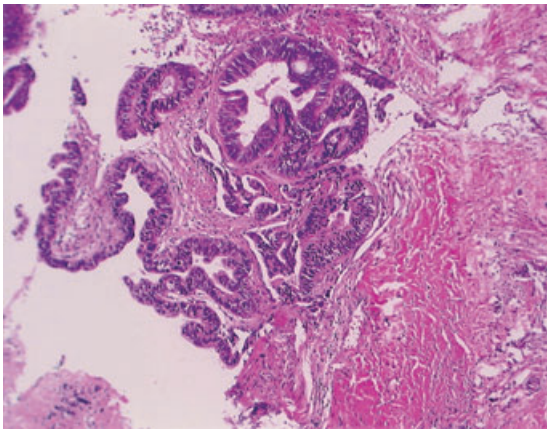

Figure 5 Photomicrogragh of the biopsy specimen obtained from the right infratemporal mass lesion showing moderately differentiated mucinous adenocarcinoma. Hematoxylin and eosin, magnification ×100.

Radiographic findings of adenoid carcinoma arising in the maxillofacial region are characterized by the presence of a mass with ill-defined margin accompanied by necrotic lesion without the formation of a large cystic lesion (3). The case presented here basically coincided with these radiographic findings. Although adenocarcinoma commonly arises from salivary glands including minor glands, mucinous adenocarcinoma in the infratemporal fossa is unusual because there are no salivary glands in this area. It was reported that 40% of all tumors occurring in the salivary glands are malignant, 5.7% of these malignant tumors were diagnosed as adenocarcinoma, and half of these originated from the parotid glands (3). It was also reported that 3.3% of parotid gland malignancies were metastatic tumors and that metastasis to the maxilla is more frequent than that to the mandible (1). In addition, primary mucinous adenocarcinoma of the head and facial skin is very rare (4).

Therefore, it was considered that mucinous adenocarcinoma in this case originated from the deep lobe of the parotid gland or heterotropic salivary gland, or was a metastatic tumor from another organ. Although cases of squamous cell carcinoma and sarcoma arising from chronic osteomyelitis or chronic medial otitis were reported as tumors attributable to inflammations (5), there is no available literature describing mucinous adenocarcinoma subsequent to inflammation.

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