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Case report

The mandibular condyle as uncommon metastatic site of neuroendocrine carcinoma: Case report and review of literature

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ABSTRACT

Temporo-mandibular joint (TMJ) metastases are a very rare event and only 73 cases are reported in literature. In about 40% of cases condylar metastases represent the first clinical manifestation of a tumor of elsewhere and may then allow an early diagnosis. However, the identification of this tumoral process can be difficult as in over 50% of the cases it has a nuanced clinical presentation that is very similar to temporo-mandibular disorders.

The first case of metastatic neuroendocrine carcinoma (NEC) of the temporo-mandibular joint (TMJ) mimicking a temporo-mandibular joint disorder is presented in this report. Furthermore, an extensive review of the literature has been performed in order to establish a correct diagnostic–therapeutic protocol for these oncologic patients.

1. Introduction

Primary neoplasm of the mandible are more common than metastatic disease, which represents only 1% of such tumors [1]. Metastases are more commonly seen in the hematopoietically active marrow of the skeletal bones. The cancellous bone at these levels is indeed rich with sinusoidal vascular spaces that permit tumor cells penetration. The mandible is not a site of active marrow in humans, particularly in older individuals. When cancellous marrow is present, it is usually in the posterior aspect of the mandible [2].

If metastatic tumors of the mandible are rare, involvement of the mandibular condyles by such growths is even rarer and, since the first description by De Cholnoky in 1941 [3], only 73 cases are reported in international literature.

An unusual case of metastatic neuroendocrine carcinoma (NEC) of the temporo-mandibular joint (TMJ) is described in this report. The caecum was the site of arising of the primary tumor. This is the first report of metastatic TMJ involvement of a NEC.

2. Case report

A 66-year-old Caucasian man presented with an episode of acute intestinal obstruction. His medical history included chronic ischemic heart disease, hypercholesterolemia and hypertension. He was therefore hospitalized at the Department of Surgery.

A total body CT scan was then executed showing a 9 cm caecal mass with infiltration of the last ileal loop and the appendix. A regional lymphonodal involvement and the presence of a single liver metastasis were revealed. The patient underwent immediate right colectomy with contemporary resection of the hepatic metastasis. Histological examination revealed a Large Cell NEC of the large bowel. The intestinal mucosa was infiltrated by a proliferation of tumor cell faintly arranged in a organoid growth pattern. The tumor was composed of large cells layers with scant cytoplasm and enlarged, pleomorphic nuclei. Numerous apoptotic bodies and mitotic figures were observed. High power magnification of the tumor showed glandular differentiation and prominent intracytoplasmic mucin vacuoles, yielding a "signet ring cell" appearance. Tumor cells showed immunoreactivity for chromogranin and Ki-67; labeling index was 95% (Fig. 1).

☆ AsianAOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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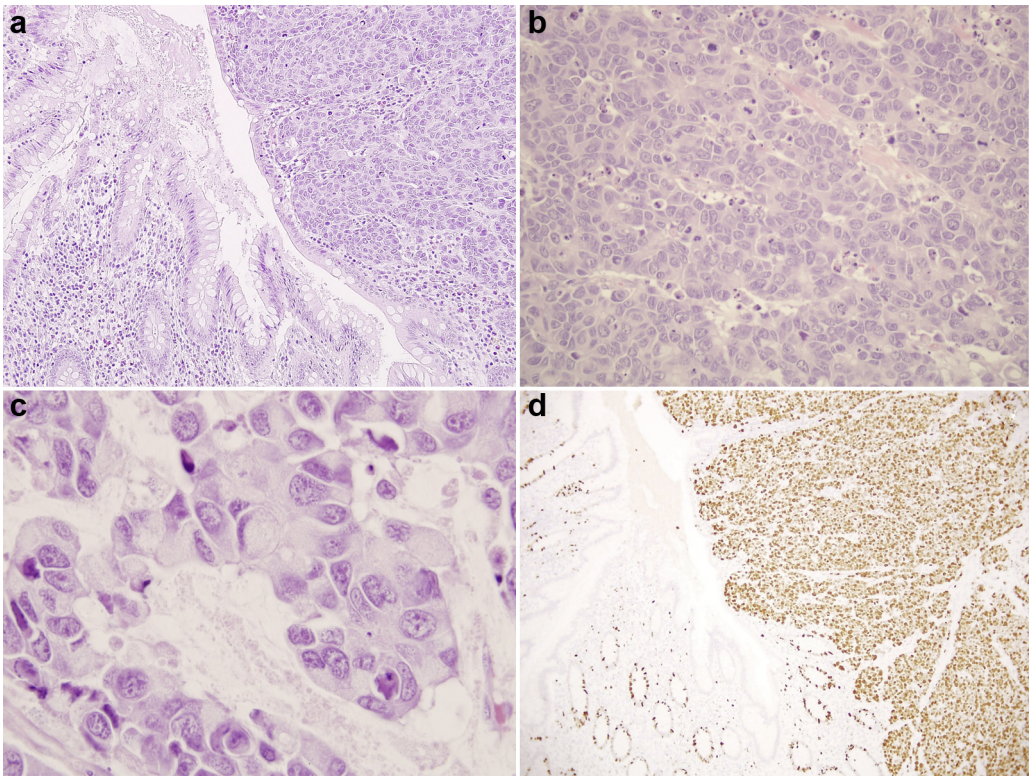

Fig. 1. Histologic features of caecum NEC. A (H&E, 20×): The intestinal mucosa is infiltrated by a proliferation of tumor cell faintly arranged in a organoid pattern. B (H&E, 40×): The tumor is composed of sheets of large cells with scant cytoplasm and enlarged, pleomorphic nuclei. Numerous apoptotic bodies and mitotic figures are observed. C (H&E, 100×): High-power magnification of the tumor showing glandular differentiation and prominent intracytoplasmic mucin vacuoles of tumors cells, yielding a "signet ring cell" appearance. D (Peroxidase stain, 40×): The tumor cells show immunoreactivity for chromogranin. Ki-67 labeling index was 95%.

During the post-operative period the patient complained ingravescent right TMJ pain and was referred to Maxillo-Facial Surgery Department for evaluation.

He reported that, actually, right TMJ pain and limitation of jaw movements started about 8 months before. For that problem he already turned to his dentist that, in the suspicion of a TMJ disorder, prescribed NSAIDs and myorelaxant therapy. Two weeks after, due to the symptoms persistence, the patient performed radiological exams. Orthopantomograph and dynamic TMJ radiographs were totally negative. TMJ magnetic resonance, of which the patient had no images, referred anterior displacement of the right disk, without reduction, intra-articular effusion and morphological alterations of the condyle compatible with arthritic degeneration. Clinical and radiological diagnosis of not reducible TMJ disk anterior displacement was then made and the patient begins a conservative therapy with occlusal bite that was continued, without any benefit, for 5 months until the admission at the Department of Surgery.

Clinical examination of the patient did not show masses or swelling of the right TMJ, masticatory muscles appeared contracted and painful. The patient complained pain both at rest (VAS 5) and during mandibular movement (VAS 9). Maximum mouth opening was 15 mm with right deviation; left lateral excursion was 2 mm whereas there was no restriction of the right lateral excursion. Parotid glands secretion was clear and the patient did not show cervical lymphadenopathy. The oral cavity inspection was normal. There was deep bite, Class II occlusion with complete molar edentulism.

To investigate the presence of a TMJ metastatic lesion, maxillofacial contrasted CT scan was then performed showing structural subversion of the right condyle with osteosclerotic areas alternate to 3–5 mm in diameter osteolytic lesions. The periosteum and

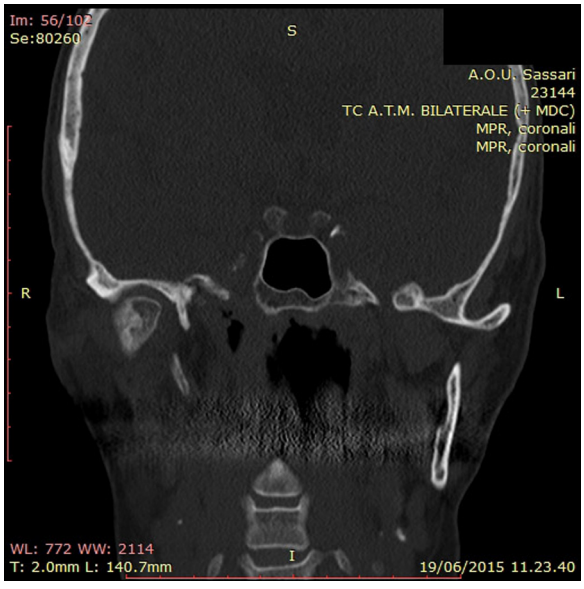

Fig. 2. Right TMJ CT-scan showing osteosclerotic areas alternate to 3–5 mm in diameter osteolytic lesions.

the lateral pterygoid muscle presented increased thickness and oedema without others significant alterations (Fig. 2).

The patient was submitted to open biopsy, frozen sections confirmed the malignancy suspicion. Condylectomy with healthy margins was then performed in the same surgery (Fig. 3). Definitive histologic examination confirmed the diagnosis of metastatic lesion showing epithelial scattered signet-ring cells containing intracytoplasmic mucina and poorly formed glandular lumen arranged in

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