



CASE REPORT

Intraosseous mucoepidermoid carcinoma

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Abstract

Mucoepidermoid carcinoma (MEC) is a common malignant tumor composed of mucinous cells, clear cells and squamoid tumor cells. It is usually localized on the salivary glands, however can very rarely be intraosseous. This tumor affects women more than men and is more common in the mandible. In this report, we present a case of a 45-year-old woman with a painless intraoral mass for 10 years which was repeatedly misdiagnosed, evolving into an extensive lesion. After an appropriate approach, she was diagnosed with MEC of the mandible and submitted to a left hemimandibulectomy accompanied by microsurgical reconstruction with excellent results.

Keywords: bone neoplasms; head and neck neoplasms; mandibular reconstruction; microsurgery.

Introduction

Mucoepidermoid carcinoma (MEC) is a malignant tumor composed of mucinous cells, clear cells and squamoid tumor cells forming cystic and solid patterns¹. It is the most common malignant tumor of the salivary glands, mainly seen in the parotid gland¹. It occurs mostly in women, with a greater prevalence at 40-50 years old². An intraosseous MEC, also known as central MEC, is a very rare condition, accounting for 2-3% of all MECs reported³. This type of MEC occurs more often in the mandible, especially the posterior part³. In this report, we present a case of a 45-year-old woman with MEC of the mandible with excellent results after treatment.

Case report

A 45-year-old woman presented was referred to the University Hospital of Federal University of Ceara (UFC) Medical School complaining of a painless intraoral mass on the posterior part of left mandible, which was growing for 10 years. During this period, the patient had been repeatedly diagnosed and treated as gingivitis. At the examination, a left retromandibular lesion of approximately 5 cm was observed, associated with bone loss in the buccal and lingual bone plates. Computed tomography showed a lytic and insufflated lesion at the angle and left branch of the mandible with 3.0 x 2 cm, without erosion of the cortical bone. An incisional biopsy of the lesion was performed and the result was compatible with Oncocytic Mucoepidermoid Carcinoma.

Left hemimandibulectomy was performed with supraomoid neck dissection and microsurgical reconstruction using a free flap of the fibula of the right

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lower limb (Figure 1 and 2). Histopathological report showed a low grade mucoepidermoid carcinoma with extensive bone involvement of the underlying jaw. The adjacent and deep margins, as well as the lymph nodes, were free of neoplasia. However, perineural infiltration was present and radiotherapy was indicated. The patient presented good evolution in the immediate and late postoperative period. Presents very satisfied with aesthetic results and without complain within 6 months of follow-up (Figure 3).

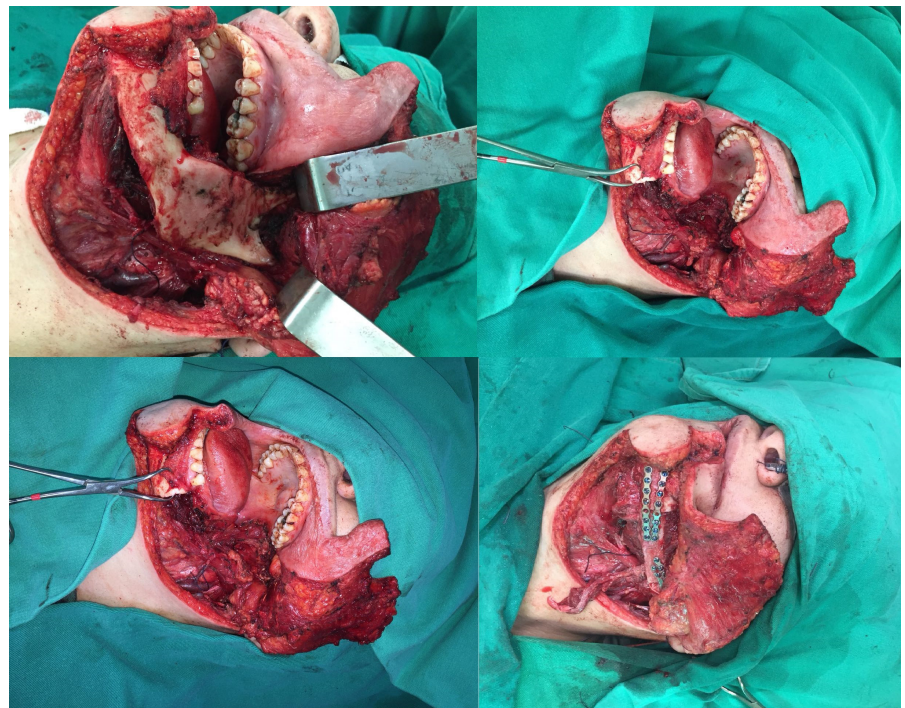


Figure 1. Intraoperative images.

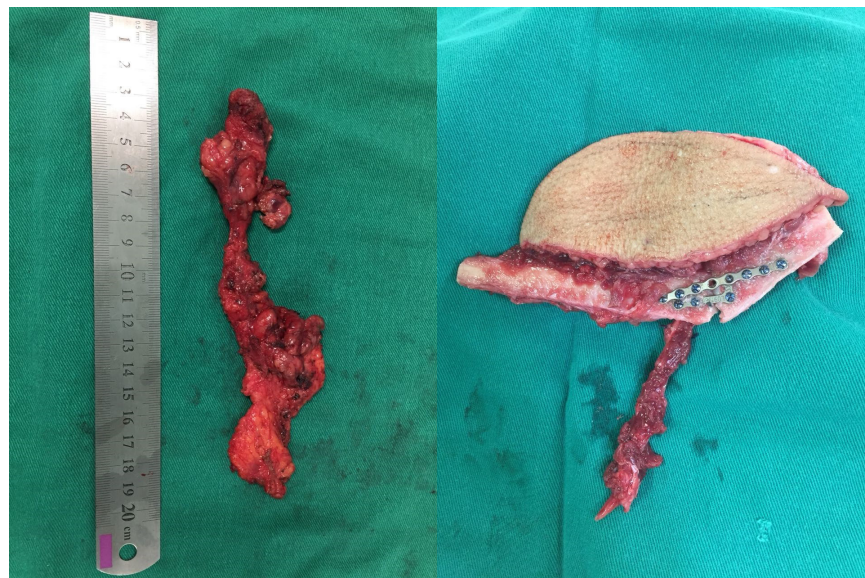


Figure 2. Surgical specimens.



Figure 3. Patient with 6 months postoperative.

Discussion

MEC represents one third of all salivary malignancies³, affecting mostly the parotid gland (86%)⁴, but there are few cases of intraosseous MEC. Central MEC was first reported in 1939 and there are about 200 cases in literature so far². It is rare but is a well-known condition⁴. It affects jaw bones, affecting the mandible 3 times more than the maxilla². The most common site of occurrence is the premolar-molar-angle region of the mandible, associated with mandibular cysts in almost half of the cases⁴. Intraosseous MEC involves woman twice as frequently as men². The symptoms may include painless swelling, pain, paresthesia, numbness and loose teeth¹.

Initial evaluation frequently includes radiographic images; however a biopsy is essential to the diagnosis⁵. Radiographic features includes a well-circumscribed unilocular or multilocular areas of radiolucency². A classification based on the radiographic characteristics of the tumor was proposed¹. Stage 1 is the lesion with an intact cortex layer and without evidence bone expansions¹. A stage 2 lesion is when there is expansion to the bone but do not disturb the integrity of the cortical¹. The stage 3 is characterized with disruption of the periosteum integrity or cortical perforation or any mass with nodal involvement¹. These stages are used for determining the prognosis, being the stage 3 the poorest prognosis¹. However, these clinical presentation and radiographic features could represent other osteolytic and odontogenic processes, so a biopsy is always indicated².

Fine needle aspiration and incisional or punch biopsies are hard to do in this type of tumor¹. Open biopsies through the bones are often necessary¹.

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Histopathological classification includes low grade, intermediate and high grade tumors^{3,5}. Highly differentiated lesions with preponderance of macro- and microcysts are categorized as low grade³. Lesions with intermediate grade have predominance of intermediate cells and a few cysts, associated with islands of epidermoid cells and mucin-producing cells³. Poorly differentiated tissues with prevalence of intermediate and epidermoid cells are high grade³.

There are well-established criteria for the diagnosis of intraosseous MEC: (1) exclusion of an odontogenic tumor, (2) exclusion of another primary tumor whose metastasis could histologically mimic a central tumour, (3) radiological evidence of bone destruction, (4) intact cortical plates on CT, (5) histopathological confirmation, and (6) detectable intracellular mucin⁴.

The treatment is essentially surgical, with en bloc resection, even for low-grade tumors⁵. Segmental resection of the jaw may be necessary⁵. In the present case, a hemimandibulectomy was performed, with a fibula free flap reconstruction. Excellent aesthetic and functional results were obtained. Other conservative therapies, such as curettage or enucleation, are not recommended since they have a higher percentage of recurrence: 40% of the cases had recurrence of the disease after conservative therapy compared to only 4% of the cases in more aggressive treatment². Neck dissection is recommended for patients with high grade tumors or with regional lymph node metastasis¹. Radiotherapy is usually indicated for high-grade tumors and in cases with close surgical margins⁵. In our case, it was indicated because of the perineural infiltration.

The prognosis of intraosseous MEC is not easy to determine, but it is established to be good⁵. The 5-year survival rates for low grade lesions are up to 95% and for high-grade tumors are 40%¹. Metastasis occurs in about 9% of the cases, mainly to regional lymph nodes, ipsilateral clavicle, lungs, and brain². The patients must be followed for a longer period up to 10 years⁵, due to high rates of local recurrence and late distant metastasis⁵.

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