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Case Report

Central mucoepidermoid carcinoma in a young patient: A case report and review of the literature

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ABSTRACT

Background: Central mucoepidermoid carcinoma is a rare malignant salivary neoplasm arising inside the bone of the jaws. We report a rare case of central mucoepidermoid carcinoma of a 16-year-old girl arising in the maxilla. Clinical and radiological analyses were shown and discussed, as well as the surgical treatment.
Methods: A left maxillectomy from the last molar to the first premolar was performed, and the site was reconstructed using a fibula free flap stabilized with titanium plates.
Results: The young patient was followed up for more than 6 years, and to date no recurrence was observed. A literature review of the 11 rare previous cases was provided to guide the clinician in the diagnosis and management of this unusual glandular tumor.
Conclusion: It is of immense importance to differentiate the central mucoepidermoid from other osteolytic lesion and odontogenic cysts because of his malignancy and local aggressiveness. The treatment option is the radical excision, with the evaluation of neck nodes, and radiotherapy is only recommended in the most aggressive of cases.

1. Introduction

Mucoepidermoid carcinoma is the most frequent malignant neoplasm of the salivary glands, effecting those with a mean age of 45 years old [1]. It occurs frequently in major salivary glands, especially the parotid, and in salivary glands of the palate and buccal mucosa [1]. The jaw's cortical bone is sometimes involved *ab estrinseco* by mucoepidermoid carcinoma where the tumor makes contact with the bone. On rare occasions, mucoepidermoid carcinoma can be identified as a total intra-bony lesion without soft tissue involvement. This unusual pathological entity is called central mucoepidermoid carcinoma (CMC). It is a very rare variant of mucoepidermoid carcinoma representing 2–4% of all cases/occurrences [2,3], and occurs in the fourth and fifth decade, especially in the maxilla of women [4]. However, CMC has also been reported in young patients (less than 18 years old). The pathogenesis, treatment and prognosis of CMC are still controversial due to its rarity. This case report describes a CMC arising in the maxilla of a 16-year-old female patient diagnosed in December 2007 and surgically treated in January 2008 at the Maxillo-Facial Unit of the S. Orsola-Malpighi Hospital of the University of Bologna, Italy. The patient was followed up for 6 years. The 11 previous cases of CMC arising in young patients (less than 18 years old) reported in the literature from 1952 to 2014 are described, summarizing the main characteristics of this rare malignant variant of mucoepidermoid carcinoma [5–14].

2. Case report

On December 2007, a 16-year-old white female patient came to our attention due to an occasional radiological finding of left maxilla lesion. The young patient did not report any symptoms. There was neither precedent toothache nor trauma. Clinical examination revealed the presence of a swelling in the palate that was painless and the overlying buccal mucosa was intact (Fig. 1). The expansive central lesion caused the swelling of the mucosa. The mucosa was

☆ 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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intact with blue-red color mimic a mucocele or a vascular lesion. Neck evaluation was negative for lymph-node involvement. The panoramic radiograph revealed a well-defined radiolucent lesion close to the premolars and first molar roots (Fig. 2).

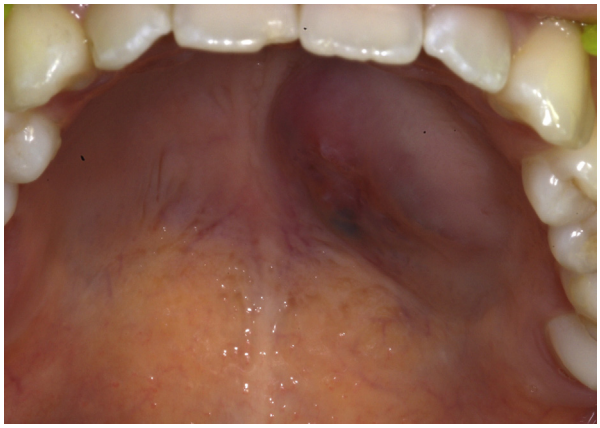

Fig. 1. Intraoral swelling of the palate caused by the expansion of the central lesion.

The computed-tomography (CT) scan revealed an expansive multilocular intra-bony radiolucency area of the left maxilla, without extra-bony spread (Figs. 3 and 4). Based on clinical and radiological features the differential diagnosis could be an odontogenic cyst, fibrosseous lesions or a keratocystic odontogenic tumor.

An incisional biopsy was performed resulting in the diagnosis of a mucoepidermoid carcinoma.

A left maxillectomy from the last molar to the first premolar was performed (Fig. 5). The specimen consisted of a firm and solid mass, with cystic spaces and no necrotic areas. The radical excision of the tumor was confirmed by the pathologist. The surgical gap was simultaneously reconstructed using an osteomuscolar vascularized fibula free flap stabilized with titanium plates (Fig. 6). The muscular component of the flap was used to reconstruct the oral mucosal layer. Gradual and complete mucosal metaplasia was recorded in the intra-oral component of the flap during the follow-up. Fig. 7 shows the photomicrographs stained with hematoxylin and eosin at low magnification. Microscopically, the specimen was composed of mucous and epidermoid cells in a fibrous connective stroma and bone. Cells show positive mucin staining, and it was excluded a histopathologically similar primary lesion of salivary glands.

The definitive diagnosis was central mucoepidermoid carcinoma at intermediate grade of malignancy. Two years after

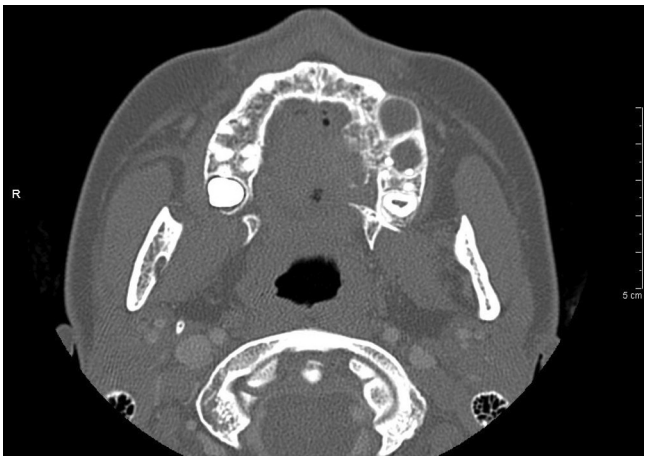

Fig. 3. Computed-tomography scan showing a multilocular radiolucency area of the left maxilla.

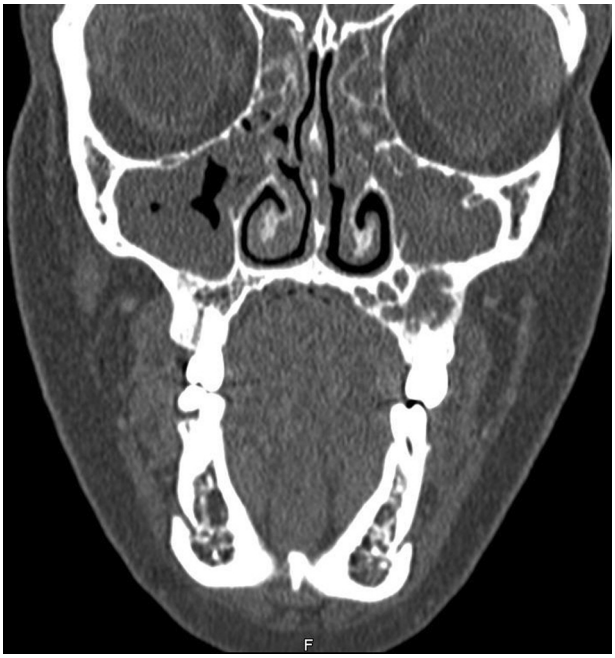

Fig. 4. Coronal CT scan revealing the tumor inside the maxillary bone without extra-cortical spread.

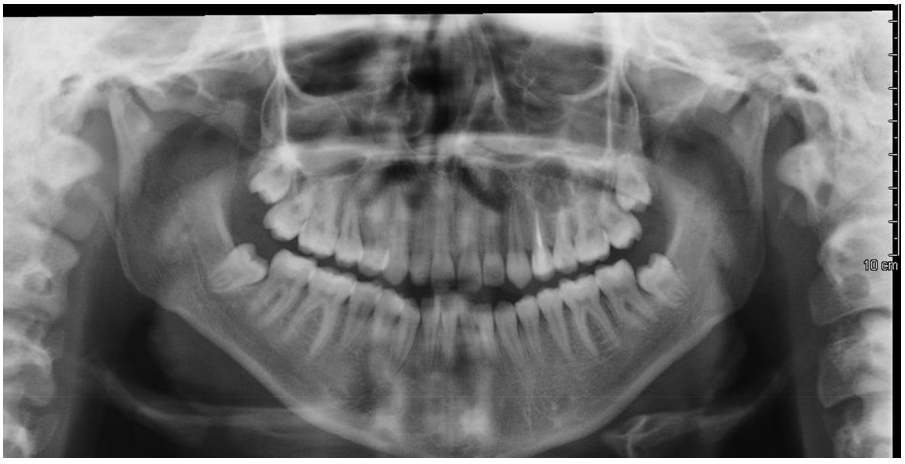

Fig. 2. Panoramic radiograph showing a well-defined radiolucent area in the left maxilla.