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Case Report

## Central Mucoepidermoid Carcinoma of the mandible—From a histopathologic perspective



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#### ABSTRACT

Salivary gland tumors occurring intraosseously within the jaws are rare. The Central Muco Epidermoid Carcinoma (CMEC) is the most common salivary gland tumor to occur in an intraosseous location. The histogenesis of this tumor is controversial. This paper aims to highlight the rare histopathologic findings in a case of CMEC of the mandible, with a long term follow up, which throws some insight into the origin of this tumor based on the proposed theories of its origin.

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#### 1. Introduction

Salivary gland tumors occurring intraosseously within the jaws are rare lesions. There is a general consensus that mucoepidermoid carcinoma is the most common salivary gland tumor found in an intraosseous location. Central Muco Epidermoid Carcinoma (CMEC) occurs about three times more commonly in the mandible than maxilla. The histogenesis and biologic behavior of this tumor is controversial. This paper aims to highlight the histogenesis of CMEC of the mandible, with reference to its theories of origin based on rare histopathologic findings, in a case diagnosed as CMEC of the mandible.

#### 2. Case report

At the time of reporting in June 2001 to one of the maxillofacial centers in Northern Kerala, this female patient was 52 years old with a complaint of swelling in the right side of her lower jaw. She had given a history of extraction of her right lower molars and cyst enucleation, performed at the same center three years back, only to

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notice a recurrence of the swelling one year after the initial surgery. The initial histopathologic report was suggestive of a keratocystic odontogenic tumor.

#### 2.1. Examination

The recurrent swelling was diffuse and bony hard and extended anteroposteriorly from the right second premolar region to the ramus of the mandible with buccal cortical bone expansion. She had no other symptoms of pain or parasthesia. The cervical lymph nodes were not enlarged. There was no evidence of regional or distant metastasis. The covering skin and mucosa were intact.

#### 2.2. Radiographs

CT examination showed an expansile lytic lesion involving the same region. The bony margins were irregular and the buccal cortex was perforated (Fig. 1). Orthopantomograph revealed multilocular radiolucency involving the body and ramus region of mandible (Fig. 2).

#### 2.3. Management and follow up

Based on the incision biopsy report the lesion was excised. A hemimandibulectomy on the right side was performed. The patient also received adjuvant radiotherapy and chemotherapy. The patient was followed up at regular intervals with no clinical or radiographic evidence of recurrence. A bony reconstruction was

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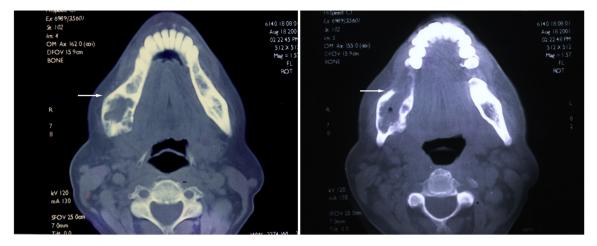


Fig. 1. Preoperative CT scan showing expansion and perforation of the buccal cortex.



**Fig. 2.** Preoperative orthopantomograph showing multilocular radiolucency involving the body and ramus region of mandible.

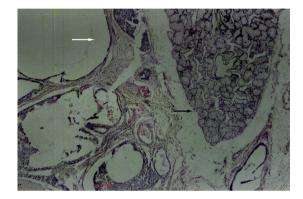
not carried out due to patient non-compliance. During the course of follow up, patient had extracted all her remaining lower teeth and is presently on a lower denture that is soft tissue, bone and implant supported (Fig. 3). The patient at 11 years follow up is doing well, with no evidence of local recurrence, regional or distant metastasis except that she is lethargic due to the aging process.

#### 2.4. Histology

The microscopic examination of the resected bony specimen revealed features of low grade mucoepidermoid carcinoma. The neoplasm comprised predominantly of cystic spaces containing mucous and lined by numerous mucous secreting cells (Figs. 4 and 5). Nests of squamous cells formed the epidermoid component in a delicate fibrous stroma (Fig. 5). Some sections showed lobules of normal salivary gland tissue (Fig. 4). Areas of odontogenic epithelium in interlacing strands with areas of mucous metaplasia were seen in some sections (Fig. 6). The histopathologic diagnosis of a low grade CMEC was made. It is considered as a rare histopathologic finding because; the aberrant normal mucous salivary gland tissue and the odontogenic epithelium undergoing



Fig. 3. 11 years post operative radiograph showing healthy resected bone margins.

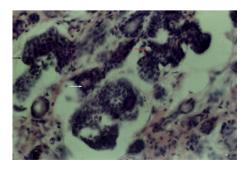


**Fig. 4.** Large cystic spaces (white arrow) intermingled with islands of epidermoid cells (red arrow). Normal mucous salivary gland tissue (black arrow) also seen (original magnification  $40\times$ ). (For interpretation of the references to color in this figure legend, the reader is referred to the web version of the article.)

mucous metaplasia are visualized in various sections, thus validating the theories of origin of this tumor.

#### 3. Discussion

CMEC of the jaws is a very rare lesion which represents 2–4% of all mucoepidermoid carcinomas [1,2]. The most frequent age group affected falls in 40–59 years with a female preponderance of 1:1.45. The lower jaw is affected in 82% of the cases with 83% of the cases occurring in the molar region, which is the most probable location of developing dentigerous cysts and odontogenic tumors [3,4]. The most likely source of CMEC is the neoplastic transformation of the



**Fig. 5.** Solid clusters of proliferating epidermoid cells (black arrows), mucous cells (white arrows) along with ductal proliferation (red arrow) (original magnification  $40\times$ ). (For interpretation of the references to color in this figure legend, the reader is referred to the web version of the article.)

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