CENTRAL MUCOEPIDERMOID CARCINOMA OF THE MANDIBLE: A CASE REPORT

Adjei J.R[1], Frimpong P.B[1], Nsiah A.O[1] , Gyau-Darko B[2] and Nartey N.O[1]

[1] Department of Oral Medicine and Oral Pathology, University of Ghana Dental School
[2] Department of Oral and Maxillofacial surgery, 37 Military Hospital.

Correspondence author: Adjei JR, Department of Oral Medicine and Oral Pathology, University of Ghana Dental School.

Correspondence e-mail: jonnierandal@gmail.com

DOI: https://dx.doi.org/10.4314/gdj.v20i1.8

ABSTRACT

Mucoepidermoid carcinoma is the most common malignant tumour of the salivary glands. It usually occurs in the parotid glands. Primary intraosseous mucoepidermoid carcinoma is rare, making up 2-3% of all mucoepidermoid carcinoma. They usually affect the mandible, with the common clinical presentation being jaw swelling. We report a case of a primary intraosseous mucoepidermoid carcinoma in a 48-year-old African male, who was managed surgically by left hemimandibulectomy, on account of a provisional diagnosis of ameloblastoma.

Keywords: Intraosseous, Mucoepidermoid, Carcinoma

INTRODUCTION

Mucoepidermoid carcinoma (MEC) is the most prevalent malignancy of the salivary glands. It accounts for about 30% of all malignant salivary gland lesions, and 12% of all salivary gland tumours, with the parotid gland being the most common site, about 60-90% for the carcinoma[1,2]. The palate happens to be the most common site for minor salivary glands involvement; accounting for about 41.1%[3].

The World Health Organisation (WHO) describes MEC as a distinct salivary gland malignancy which is composed of mucinous, intermediate (clear cells) and squamoid tumour cells which may form cystic or solid patterns surrounded by dense fibrous-connective tissue[4].

Primary intraosseous (central) mucoepidermoid carcinomas are rare; making up about 2-3% of all mucoepidermoid carcinomas[5], with about 80% occurring in the mandible[6].
The pathogenesis of central mucoepidermoid carcinoma is controversial and several theories have been proposed, including; i) neoplastic changes of a retromolar mucous gland which was entrapped in the mandible during development; ii) neoplastic changes of mucous secreting cells in an odontogenic epithelium; iii) neoplastic transformation of the lining of a maxillary sinus that has been entrapped within the mandible[3].

There are few cases of central mucoepidermoid carcinoma reported in the English literature, about 130 cases[3,7]. We hereby present a new case of central mucoepidermoid carcinoma in a 48-year-old African male.

CASE REPORT.

A 48-year-old diabetic male presented to the Maxillofacial Unit at the 37 Military Hospital, Accra, Ghana, in September 2018 with chief complaints of swelling and pain of the left mandible for year's duration. The patient noticed the swelling two weeks after tooth #37 and #38 extraction at another hospital. The swelling has increased in size over the period.

There was facial asymmetry. The swelling was situated at the pre-auricular region, measuring 6x3cm in diameter. The mass was not attached to the overlying skin and underlying tissues, and there was no differential warmth. There was no cervical lymphadenopathy.
Intraorally, the diffuse bony-hard swelling involved the body of the mandible. The buccal and lingual cortices were expanded with inflamed overlying mucosa. There were missing 46, 37 and 38. Dark brown fluid was aspirated from the lesion.

Panoramic radiograph (figure 1) and CT scan (figure 2) both showed an extensive lytic lesion of the left mandible which extended posteriorly from the distal surface of the left mandibular first molar tooth to the ascending ramus, coronoid process and head of the condyle.

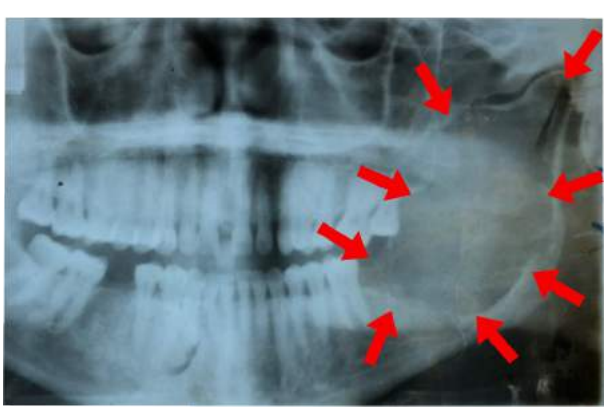

Figure 1: OPG showing unicystic radiolucent lesion of the left posterior mandible

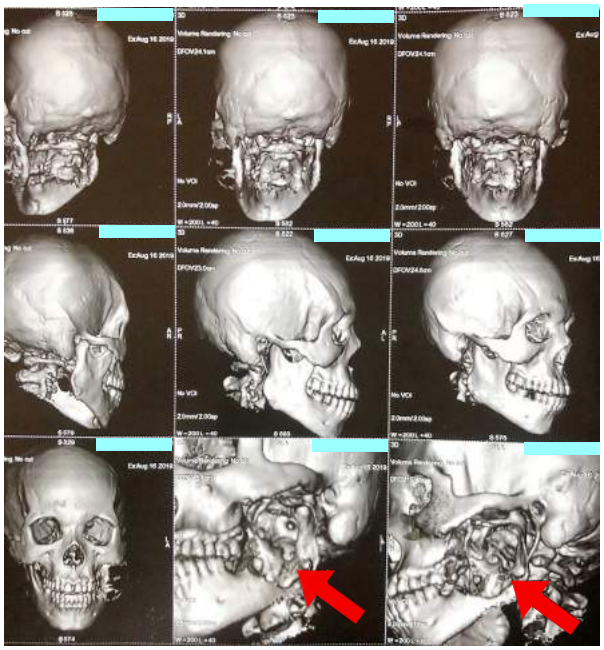

Figure 2: 3D CT Scan showing osteolysis of left mandible.

A left hemi-mandibulectomy with disarticulation was performed with a provisional diagnosis of ameloblastoma. The resected tissue was submitted to the oral pathology laboratory of the University of Ghana Dental School for histopathological examination.

The gross examination of the resected specimen revealed a dark tan soft to bony hard mass expanding both cortices and involving the mandible's ramus, coronoid process, and body.

Microscopic examination revealed a neoplasm composed predominantly of cystic spaces and an epidermoid component in a fibrous stroma. The cystic spaces were of varying sizes, lined by mucous-secreting cells (Figure 3) and cells of intermediate type. Nests and strands of squamous cells formed the epidermoid component (Figure 4). Mucinous material was seen within the cystic spaces and extravasating into the stroma.

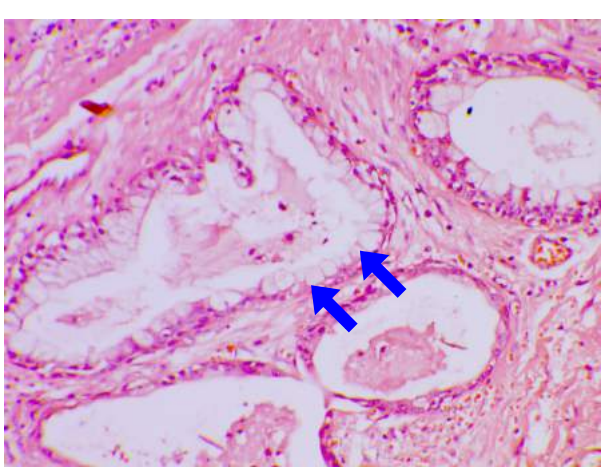

Figure 3: histologic section showing mucoid cells highlighted by blue arrows (H&E x200)

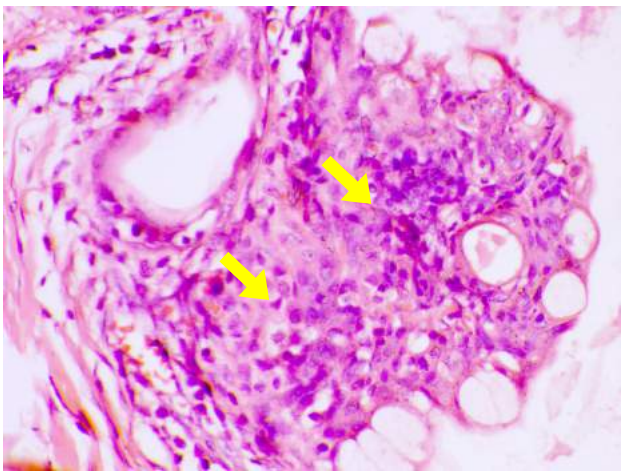

Figure 4: histologic section showing epidermoid cells highlighted by yellow arrow (H&E x200)

A definitive diagnosis of Central Mucoepidermoid carcinoma with intermediate grade was made. Surgeons were appropriately advised to follow up on the patient.

DISCUSSION.

Central mucoepidermoid carcinoma is a rare lesion first documented and reported by Lepp in 1939 in the mandible of a 66-year-old female[8,9]. Central mucoepidermoid carcinoma has a male-to-female ratio of almost 2:110. About 80% of central mucoepidermoid carcinoma occur in the mandible; however, the few that occur in the maxilla have worse prognosis due to involvement of vital organs[6,11]. The most common site of occurrence in the mandible is the posterior mandible[12], predominantly in the premolar-molar-angle region[3]. The majority of cases occur between the 4th and 5th decades of life[9], though it has been reported in all ages[5]. The usual presentation of the lesion is facial swelling caused by expansion of the bone with the associated facial asymmetry[13]. Other clinical presentations may be a pain, trismus, numbness, loose teeth, and fistula formation[7, 12]. Radiographically, there is evidence of destruction of the bone[7]. Lesions may be seen as unilocular or multilocular radiolucencies, with 50% of cases having an impacted tooth[5,12]. The case we are presenting is consistent with most of the findings in the literature.

The diagnosis of central mucoepidermoid carcinoma is not based on only histologic features but a combination of the clinical, radiographic, and histologic features, as the histologic features may look just like soft tissue, salivary gland mucoepidermoid carcinoma[7].

Alexander et al. proposed a criterion that has undergone review by other authors; Browand and Waldron; Silvergade and Schultz[5,7,12] and currently stands as (i) cortical preservation of bone; however, destruction of the cortical bone does not exclude the diagnosis, (ii) evidence of osteolysis on X-ray, (iii) exclusion of a metastatic tumour on histology (iv) exclusion of odontogenic lesion and (v) histopathological and immunohistochemical confirmation[7].

Our patient gave a prior history of extraction in the same quadrant of the jaw as the tumour after fracturing his tooth during a meal. There was no odontogenic lesion of significance, nor was there a primary salivary gland lesion. There was evidence of osteolysis and preservation of some of the cortical bone on the panoramic radiograph and the CT scan. The histopathological presentation was also typical of the lesion.

Features on the radiograph are diverse and not pathognomonic of central mucoepidermoid carcinoma. They may present as the usual cystic lesions or odontogenic tumours such as ameloblastoma[14]. Thus, it wasn't far-fetched that an initial diagnosis of ameloblastoma by the surgeons was also made in our case.

However, there is a probability that such presentations of lesions mimicking benign odontogenic tumours or cysts may be malignant lesions such as mucoepidermoid carcinoma, and, thus, prompt and proper treatment modalities will go a long way to help the patient.

Histological grading of mucoepidermoid carcinoma is based on the amount of cyst formation, cellular atypia, and the relative amount of epidermoid, intermediate, and mucous cells[15]. Low-grade lesions have high cystic spaces and mucous cells, whiles high-grade lesions have a high level of cellular atypia and more epidermoid cells[15]. In our case, the disease was graded as an intermediate based on the considerable amount of both epidermoid and mucous cells; and the few cystic spaces are seen.

Staging of mucoepidermoid carcinoma currently doesn't have a uniform system, as in the case of other lesions, such as squamous cell carcinoma, where the TNM staging system is used[2].

Radical surgery; en bloc resection with clear margins is the treatment of choice[3,12], as a review of 64 cases by Brookstone and Huvos proved 40% recurrence rate in cases where conservative surgical procedures were done and 4% recurrence rate in cases where radical surgery was done[3].

The survival rate for high-grade mucoepidermoid carcinomas is 24%; intermediate mucoepidermoid carcinoma is 83%, while that of low-grade, under which central mucoepidermoid carcinomas usually fall[5,11], is 92%[15].

CONCLUSION.

Central mucoepidermoid carcinoma is a rare entity. It mimics odontogenic lesions clinically and radiographically, making clinical diagnosis challenging, as seen in our case, where an initial diagnosis of benign ameloblastoma was made, but turned out as a mucoepidermoid carcinoma. Histologically, central mucoepidermoid carcinoma may present as a soft tissue salivary gland tumour; hence, a thorough knowledge of the clinical, radiographic presentation and understanding of the histopathogenesis of this lesion by clinicians, which have been documented as criteria for diagnosing central mucoepidermoid carcinoma, are needed to arrive at a definitive diagnosis and manage patients appropriately. This goes on to buttress the point that clinicians should incooperate their clinical, radiographic, and histological evaluations for appropriate definitive diagnosis and management.

The prognosis of the lesion is dependent on the histological grade, clinical stage, and adequate surgical resection of the tumour.

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