Central mucoepidermoid carcinoma of the mandible: a case report.

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Abstract

Primary central salivary gland carcinomas of the mandible are uncommon neoplasms. Mucoepidermoid carcinoma is a distinct pathologic entity of the salivary glands and comprises 5-10% of all salivary gland tumours. Central mucoepidermoid carcinomas (CMC) are extremely rare, comprising 2-3% of all mucoepidermoid carcinomas reported. This is a case report of central mucoepidermoid carcinoma of the anterior region of the mandible in a 37 year old female patient. Histopathological findings revealed a fibrous connective tissue within which are scattered islands, sheets and cords of neoplastic epidermoid and intermediate cell clusters exhibiting cellular pleomorphism and nuclear hyperchromatism. A diagnosis of central high grade mucoepidermoid carcinoma was made. The patient was thereafter referred to a tertiary institution with facilities for radiotherapy on account of the grade of the tumour.

Keywords: Central, Mucoepidermoid Carcinoma, Mandible.

Introduction

Mucoepidermoid carcinoma as reported by Alexander et al[1] describes specific tumours of the salivary glands and comprises 5-10% of all salivary gland tumours[2-4]. In 1945, Stewart and associates described its mucous secreting and epidermal cellular elements thus establishing it as a distinct pathologic entity[5].

Central mucoepidermoid carcinomas (CMC) are extremely rare, comprising 2-3% of all mucoepidermoid carcinomas reported[6]. A review of the English literature revealed about 120 cases of mucoepidermoid carcinoma arising in mandible[2,3,7,8-11]. This report presents a case of central mucoepidermoid carcinoma of the mandible in a 37 year old female patient and the options for management.

Case Report

A 37 year old female trader presented in the clinic with an anterior mandibular swelling of seven months duration. This was preceded by pain which was dull in nature, localized and aggravated by mastication. The swelling has been rapidly increasing in size, with mobility of the associated teeth.

On extra oral examination, there was facial asymmetry due to a localized swelling in the anterior mandible extending horizontally from about the midpoint of the right body of the mandible to about two centimeter short of the midpoint of the left body of the mandible. Vertically, it extended from the lower lip to the submental area. Overlying skin appeared tense and shiny. Submandibular nodes were bilaterally palpable, firm, tender and freely mobile.

Intraorally, there is a swelling extending from the distal aspect of lower left first premolar to the lower right first molar buccolingually. Swelling also extends to the floor of the mouth on the right. It was firm to touch, with overlying mucosa erythematous with areas of ulcerations. There was mobility of the teeth in the anterior labial segment with associated occlusal derangement **(Figure 1).** A provisional diagnosis of ossifying fibroma of the mandible was made.

An incisional biopsy of the intraosseous lesion was done. Histological examination of the specimen revealed a tissue covered in an area by stratified squamous epithelium. The subepithelial tissue is composed of moderate to densely collagenized fibrous connective tissue within which are scattered islands, sheets and cords of neoplastic epidermoid and intermediate cell clusters exhibiting cellular pleomorphism and nuclear hyperchromatism

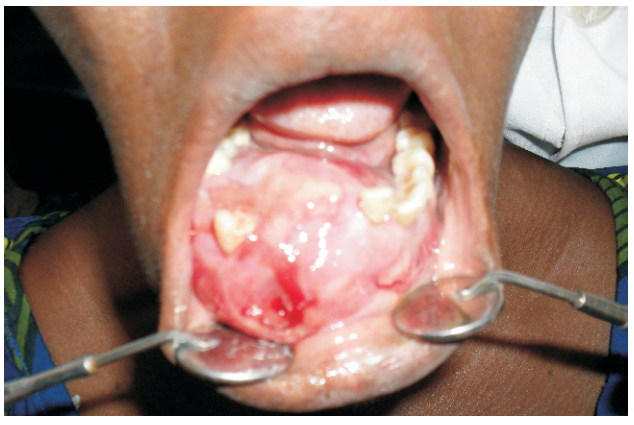

Figure 1. Clinical photograph of the central mucoepidermoid carcinoma of the mandible.

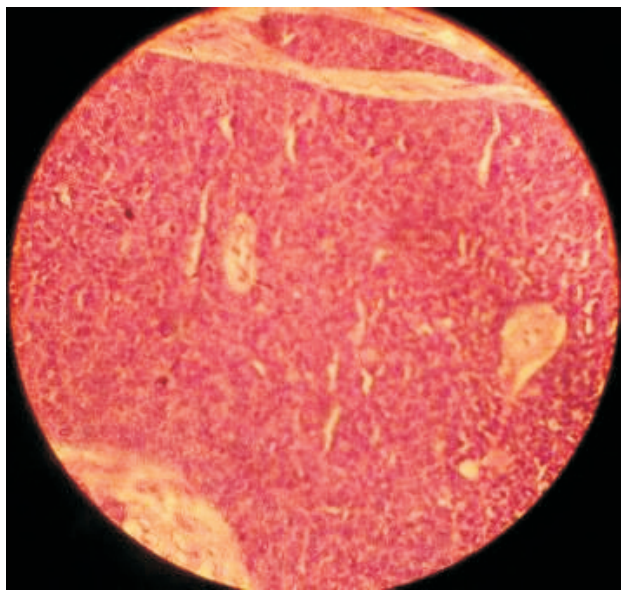

Figure 2. Photomicrograph of central high grade mucoepidermoid carcinoma showing malignant epidermoid cells and mitotic figures (H&E x 200)

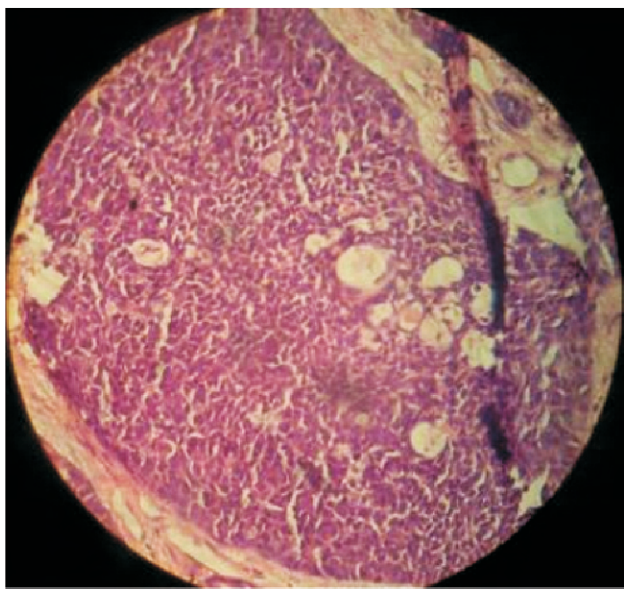

Figure 3. Photomicrograph of central high grade mucoepidermoid carcinoma showing cystic spaces containing mucin (H&E x 200)

(Figure 2). Also seen are 1 to 2 mitotic figures per high power field and occasional small cystic spaces containing mucin **(Figure 3).** There are chronic inflammatory cell infiltrate in some areas. A diagnosis of central high grade mucoepidermoid carcinoma was made. Patient was subsequently referred to another tertiary hospital with facilities for radiotherapy on account of the grade of the tumour.

Discussion

Primary central salivary gland carcinomas of the mandible are uncommon neoplasms[12]. CMC affects females twice more frequently than males and involves the mandible twice more often than maxilla[4]. The premolar-molar-angle region of the mandible has been reported as the commonest site of occurrence[3,4]. CMC has been reported in the first to seventh decade, with the highest occurrence seen in the fourth and fifth decades[4]. The gender, age and occurrence of the present case in the mandible is consistent with reports in the literature. However its location in the anterior mandible is rare.

In children, while some studies have reported equal gender ratio[4], others claim similarity to presentation in adults[13]. Due to its rare occurrence in children, it is unlikely to be a developmental disturbance or a teratoma[13]. A hormonal influence on salivary glands has been suggested as a possible aetiological factor since the tumour shows a tendency to develop at puberty[4].

The exact pathogenesis of CMC is unknown and several possibilities have been considered. These include: (1) entrapment of retromolar mucous glands within the mandible, which later undergo neoplastic transformation; (2) embryonic remnants of the mandibular and sublingual glands trapped within the mandible during development; (3) neoplastic transformation and invasion from the lining of the maxillary sinus; (4) neoplastic transformation of the mucous secreting cells commonly found in the pluripotential epithelial lining of dentigerous cysts associated with impacted third molars[3,5,14,15]. The primary location of our case in the anterior mandible supports the 2nd hypothesis.

A staging system based on the condition of the overlying bone has been proposed by Brookstones et al in 1992[7]. Lesions with intact cortical plates with no evidence of bony expansion offer the best prognosis and indicate stage I disease. Stage II disease is surrounded by intact cortical bone with some degree of bony expansion. While lesions associated with cortical perforation or nodal disease are staged III. The clinical characteristics of the present case, especially the presence of cortical perforation puts it at stage III. Metastases have been reported in 9% of central mucoepidermoid carcinomas mainly to the regional lymph nodes[7,13]. It spreads to the ipsilateral clavicle and neck occasionally.

Seifert and Sobin[16] and Auclair et al[17] highlighted certain histological criteria for the grading of mucoepidermoid carcinoma. These include: (1) low grade: highly differentiated neoplasia with a predominance of macro and microcystic spaces and presence of intermediate and mucin-producing cells; (2) intermediate grade: presence of intermediate cells and a few cystic spaces with mucin-producing cells and islands of epidermoid cells; (3) high grade: poorly differentiated neoplasia with predominance of intermediate and epidermoid cells in solid blocks and only few mucin-producing cells. The case presented here was high grade CMC.

The main stay of treatment for patients with CMC is surgery. While a high recurrence rate of about 40% has been associated with conservative surgical procedures such as curettage, enucleation, marsupialization and marginal resection with or without adjuvant radiotherapy, a recurrence rate of only about 4% has been reported in the cases treated by radical methods such as segmental resection with or without treatment of associated neck and / or adjuvant therapy.[7] Radiotherapy is recommended for high grade tumours[8,18].

Conclusion

CMC in the jaws is a rare entity. Good clinical, radiological and histological examinations are required for effective diagnosis. Close and long-term follow-up of treated cases of CMC is recommended because of the danger of late local recurrence and regional metastasis.

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