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A CLEAR CELL VARIANT OF CENTRAL MUCOEPIDERMOID CARCINOMA: A RARE CASE REPORT

Dental Science

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ABSTRACT

Central mucoepidermoid carcinoma of the jaws is a rare malignant salivary gland tumor. Here we report a case of mucoepidermoid carcinoma(MEC) of the mandible in a 70-year-old male who had a previous history of MEC of the salivary gland 40 years back. The tumor showed classical histopathological features of primary mucoepidermoid carcinoma (clear cell variant) of the mandible. On account of the early detection of this lesion it was surgically treated with no further complications

KEYWORDS

Central Mucoepidermoid Carcinoma, Clear cell variant, Calretinin, S-100, Mucicarmine, EMA.

INTRODUCTION

Mucoepidermoid carcinoma(MEC) of the salivary gland was recognized as a separate entity among salivary neoplasms by Stewart in 1945.It is thought to arise from pluripotent reserve cells of the excretory ducts of salivary gland that have the potential to differentiate into squamous, columnar and mucous cells. Ionizing radiation received during therapy has been suggested as a possible cause although no specific etiologic factors have been identified[1].Central MEC of the jaws are rare, establishing 2-3% of all testified MEC in the literature. It has been reported in the first to seventh decade, and is slightly more common in women and the posterior mandible[2].When clear cells are present predominantly in a central MEC it is known as clear cell variant. On account of the other clear cell lesions to be considered the diagnosis becomes a little more difficult[1].Here we present a rare case of clear cell variant of central MEC occurring in a patient with a previous history of MEC related to the parotid gland.

CASE HISTORY:

A 70-year-old male presented at the OPD of the Dental College, Kannur, Kerala, with complaints of pain and swelling over the lower right back teeth region since 1 & ½ years. The patient also complained of sour taste and halitosis since then.

The patient had initially noticed a small sized swelling which then gradually enlarged with mild pain,localized, continuous and progressive in nature. No aggravating factors were reported and the pain was relieved on medication. The patient also gave a history of discharge and paresthesia of the right side of the mandible since that period. No history of trauma or chronic infection was reported.

The patient reported a past medical history of MEC of right parotid gland in 1975, for which he claimed he had successfully undergone radiotherapy. However he was unable to produce records beyond a certificate relating to this .A history of radiation exposure is a known risk factor for salivary gland tumors. Patient also had a history of a cerebrovascular accident besides hypertension and diabetes and was on medication for the same since the last 10 years. Clinically no gross facial asymmetry was observable. Lymph nodes were not palpable and no clicking or deviation of the temporomandibular joint could be elicited.

Intra-oral examination showed a solitary well-defined dome shaped swelling with ulceration on the right posterior mandibular region corresponding to 47 &48 measuring approximately 4 x 1 x 1 cms in its greatest dimension with a bucco-lingual expansion of cortical plates(fig:1). The swelling extended from the first molar region to the retromolar area which partially obliterated the mucobuccal fold.

On palpation, the swelling was tender, hard in consistency, non-reducible, non-fluctuant, no pulsatile,with a mild rise in temperature. Fixity to the underlying structures was found. The surface was normal. An OPG revealed a well-defined, multi-locular radiolucency extending from posterior aspect of the body of mandible to ascending ramus sparing the condylar head.(fig 2B)

CT showed a lytic lesion involving the right body and ramus of mandible with multiple areas of cortical breach and sclerotic changes with associated soft tissue component involving the retromolar trigone area and the medial pterygoid muscle. The CT also revealed atrophic changes in the right parotid and submandibular salivary gland region with dense calcification of facial artery indicating a possibility of post radiotherapy changes(fig 2A). It also showed that the lesion was confined to the mandible.

The H and E stained section revealed an infiltrating tumor mass composed of solid and cystic tumor islands. The tumor cells arranged in sheets, glandular patterns, nests and were seen infiltrating into the surrounding tissues including bone. Fragments of necrotic bone could be seen. Clear cells with well defined borders were seen abundantly. The cystic spaces were lined by mucous secreting cells interspersed with the squamoid cells in islands. The stroma was seen to be sclerotic and showed patchy chronic inflammatory infiltrates (fig 3).

Special staining with mucicarmine confirmed the presence of mucous cells(fig 5 C).IHC staining was done using calretinin, EMA and S-100 to rule out an odontogenic origin of the tumour (fig 5 A,B,D). Following the surgical procedure the patient was recalled for follow up and advised to continue to do so for an indefinite period keeping in mind the high rate of recurrence of MEC.

DISCUSSION:

Mucoepidermoid carcinoma has been recognized as one of the most common salivary gland malignancies accounting for about 1% to 2% of all major salivary gland neoplasms and 9% of minor salivary gland tumors.Very rarely salivary gland tumors arise centrally within the jaws[3]. When a MEC lesion is composed predominantly of clear cells it should be differentiated from the clear cell odontogenic tumors. During laboratory investigation it is found that, the waterclear cytoplasm of clear cells seen in MEC is negative for mucin, fat,glycogen and lipid.MECs differentiate into three basic cell types mucin secreting ,squamoid and intermediate arranged in varying proportions[4].When the squamoid component predominates, the histological appearance is classified as a high-grade MEC . In contrast,the presence of mucin producing cells within a predominately cystic framework is regarded as low-grade MEC. [1]

They may originate from

- Entrapment of minor retro molar mucus glands which undergo neoplastic transformation
- Neoplastic transformation of mucus secreting cells in the epithelial lining of dentigerous cysts.
- Embryonic remnants of the submaxillary gland.
- Neoplastic transformation and invasion from the lining of maxillary sinus[5]

The most commonly accepted criteria for diagnosis proposed by Alexander et al., and modified by Browand and Waldron

Diagnostic Criteria :

1. Intact cortical plates (However, cortical perforation does not exclude PIOC type 4)
2. Radiographic evidence of bony destruction.
3. Exclusion of another primary tumour that is in its metastasis.
4. Exclusion of an odontogenic tumour.
5. Histopathological confirmation.
6. Detectable intracellular mucin.[5]

Based on this criteria our case was diagnosed as an central MEC. One of the characteristic features seen in the microscopy of this case was the presence of clear cells. Those seen in oral cavity are primarily of either salivary gland , odontogenic origin or secondary metastaticlesions.[6]

In the case of MEC the distinguishing feature, not seen in other clear cell tumours, is the formation of epidermoid condensations primarily found around the periphery of the tumour islands and adjacent to the vascular septa. The clear cell variant of MEC is graded in the intermediate to high grade range implying a worse prognosis than low grade tumours.[7]

Other clear cell lesions need to be considered because of the intraosseous nature of this lesion.Clear cell variant of calcifying epithelial odontogenic tumour can be distinguished by the foci of calcifications and polygonal eosinophilic cell islands.[7] A heterogeneous architecture and a rich dilated prominent sinusoidal vascular network and pronounced pleomorphism along with greater amount of hemorrhage and cytological atypia should indicate the possibility of metastatic disease like metastatic clear cell carcinoma of renal origin[8]. However these features were not evident in this lesion.

Surgery is the mainstay of treatment. In previous reports it has been suggested that treatment modalities such as radiotherapy or chemotherapy should be considered only for lesions that cannot be surgically controlled[9].The patients claim of having undergone radiotherapy for MEC 40 years back cannot be discredited despite the lack of records since the CT scan did reveal a certain degree of atrophy of the ipsilateral parotid gland which might have been brought about by the exposure to ionizing radiation.Salivary glands have been found to be reported to be sensitive to radiation, but,unlike classically radiosensitive tissues, they proliferate slowly and are made up of highly differentiated cells. The tissue's rate of proliferation correlates with the early and late effects of radiation.Late effects may occur months or years after irradiation and may be affected by vascular damage and loss of parenchymal cells[10].

These tumours usually show a good overall prognosis, but central MEC cases should be followed up for a longer period up to 10 years, due to the possibility of late recurrence or regional metastasis[10]

CONCLUSION

Primary intraosseous MECs are extremely rare tumors, usually affecting the posterior mandible as uni or multilocular irregularly bordered lesions.Clear cell variants should be treated with kid gloves on account of the higher grade.Radical surgery, adjuvant treatment and a careful histopathological evaluation of all excised tissue is important so that such neoplastic transformation may be identified and treated effectively.On account of the poor prognosis associated with MECs, early and accurate diagnosis, prompt interventions, and continued monitoring are significant.[11].Long-term follow-up is necessary, as some cases like this one would suggest,can develop late local recurrences and regional metastasis, or even a second primary lesion even after decades.

Fig 1: Intraoral swelling in the right mandible with ulceration

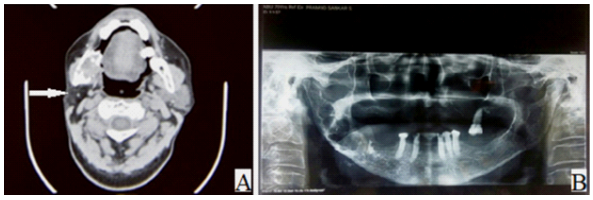

Fig 2: A) CT- Axial view – Bone window revealing significant lingual cortex destruction.(Arrow shows atrophy of the right parotid gland). B) Orthopantogram- Shows a well-defined, multilocular radiolucency extending from posterior aspect of body of mandible to ascending ramus sparing the condylar head.

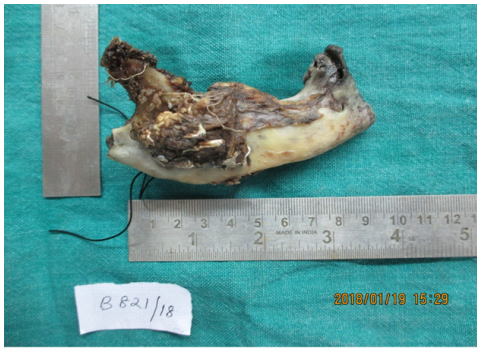

Fig 3:Right Hemi-mandibulectomy specimen with tumor.

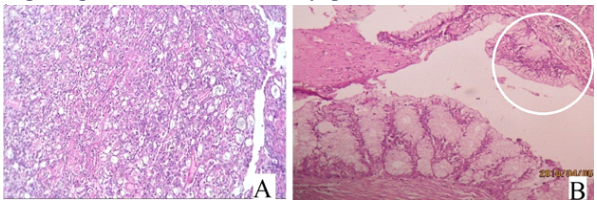

Fig 4: A) Clear cells seen in the stroma(H&E 4x)
B) Cystic space lined by mucous cells and epidermoid cells (H&E 10x)

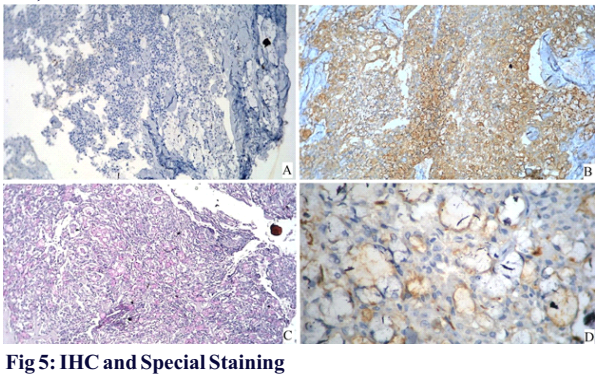

Fig 5: IHC and Special Staining
A) Negative reaction to Calretinin(10x)
B) Strongly positive for EMA(10x)
C) Special staining showing reaction to Mucicarmine(10x)
D) Immunohistochemical staining with S-100 shows focal positivity(40x)

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