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Primary Intraosseous Mucoepidermoid Carcinoma of Maxilla – A Rare Entity

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Abstract

Mucoepidermoid carcinoma is a malignant epithelial tumour of the salivary glands that contains mucous secreting, epidermoid, and intermediate cells. It is first studied and described as a separate entity by Stewart, Foote and Becker in 1945. Mucoepidermoid carcinoma represents 29–34% of malignant tumours originating in both major and minor salivary glands that exhibits diverse biological behaviour.

A primary intraosseous mucoepidermoid carcinoma was first reported by Bhaskar in 1963. He reported two cases in the mandible. Primary intraosseous mucoepidermoid carcinoma is rare malignancy accounting for 2%–4% of all MECs reported. Usually, it presents as an asymptomatic swelling. They are more common in middle-aged adults and have a slight female predilection. This carcinoma is three times more common in the mandible than in the maxilla. We hereby report a case of primary intraosseous mucoepidermoid carcinoma of posterior maxilla of left side in a 30-year-old female, who presented with palatal swelling since 8 months. This article highlights the need for considering malignant salivary gland lesions in the differential diagnosis of palatal swellings.

Keywords: Primary, Intraosseous, Mucoepidermoid Carcinoma, MEC, PIOMEC

Introduction

Salivary gland carcinoma accounts for 3 to 4% of all head and neck cancers.[1] Mucoepidermoid carcinoma (MEC) is the most common type of salivary gland malignancy.[2]

MEC occurs primarily in the major salivary glands and when it affects the minor salivary glands, it is most commonly found on the palate, followed by the retromolar region, buccal mucosa, tongue, lips and floor of the mouth, sinuses, and larynx.[1]

Primary intraosseous mucoepidermoid carcinoma is a rare neoplasm of the jaw bones. Even though theories have been proposed based on the neoplastic transformation of the epithelial mucosa of odontogenic cysts or ectopic salivary tissue.[1] It presents usually as a painless mass with a gradually increasing in size and is noticed by the patients themselves within a year or less of onset. This tumor is of clinical significance as it may metastasize, resulting in a poor prognosis.[2] This tumor is mostly misdiagnosed clinically as well as radiographically as a benign odontogenic tumor or cyst. The main modality of treatment for patients with this neoplasm is radical surgical resection, offering a greater chance of cure than the more conservative procedures, such as enucleation or curettage. The rate of local recurrence associated with conservative treatment is 40%, contrasting with a rate of 13% for the more radical treatment. Metastases have been reported in about 12% of the cases.[1]

Case Presentation :-

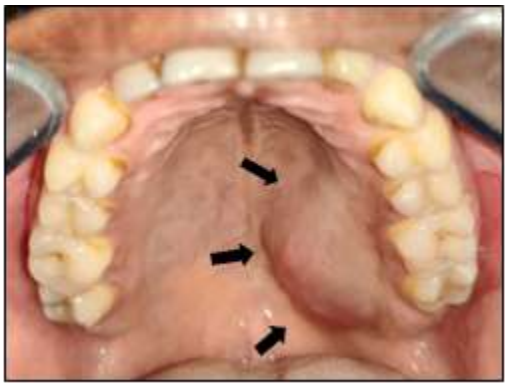
A 30-year-old female came to the department of Oral Medicine Diagnosis and Radiology with complaints of swelling on upper left palatal region since 8 month. The swelling was insidious in onset, slowly progressive and asymptomatic. There was no relevant medical or personal history.

On examination, no extraoral asymmetry seen [Fig. 1]. Intraoral examination revealed swelling of approximately oval in shape with around 2.5 cm × 2 cm size was present at left posterior palatal region, extending from premolar region to maxillary tuberosity anteroposterior and from midpalatal region to gingival margin mediolaterlly. Mucosa over swelling was slightly erythematous with punctum seen at central part of the swelling [Fig. 2]. On palpation swelling was firm, tender, with a nodular surface and fixed to underlying tissue. There was no ulceration or discharge, no absent teeth, and no cervical lymphadenopathy.

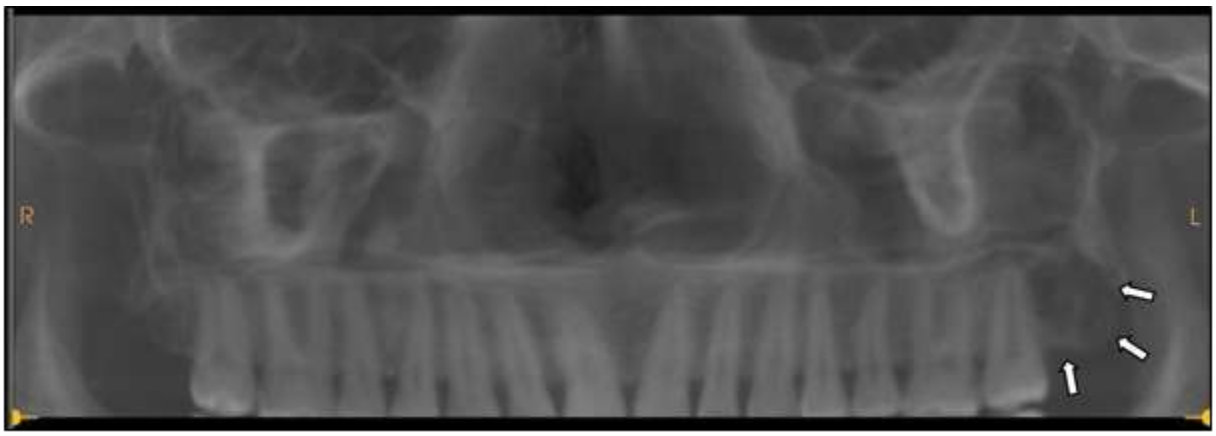
Based upon the history and clinical presentation, a provisional diagnosis of benign odontogenic tumour involving left posterior maxilla was made. Odontogenic cyst, soft tissue tumour and neoplasm of minor salivary glands was included in the differential diagnosis.

On radiographic examination, Cone beam computed tomography (CBCT) revealed a multilocular radiolucency extending from midpalatal region to buccal cortex mediolaterally and from maxillary tuberosity to posteriomost palatal region anterioposteriorly with the epicenter in the left maxilla. Also elevation of floor of maxillary sinus on left side due to lesion seen.

[Fig 1]



[Fig 2]



[Reconstructed Panoramic Image]

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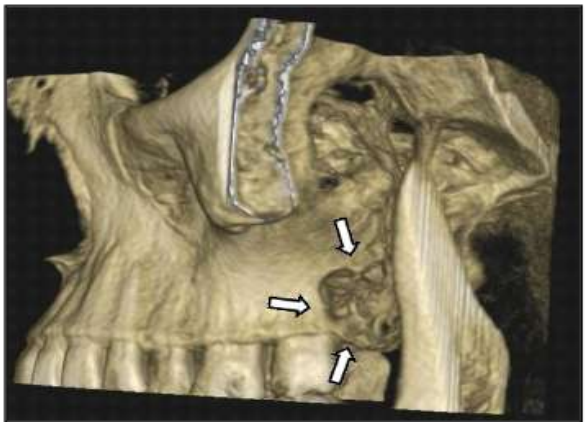
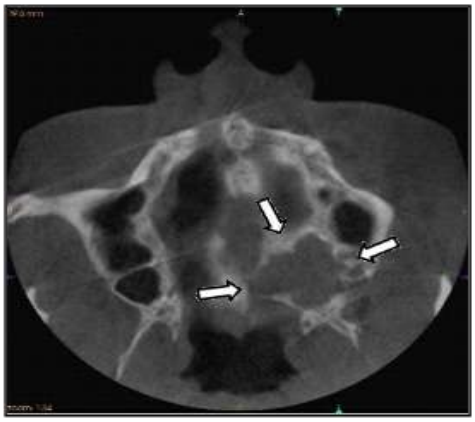
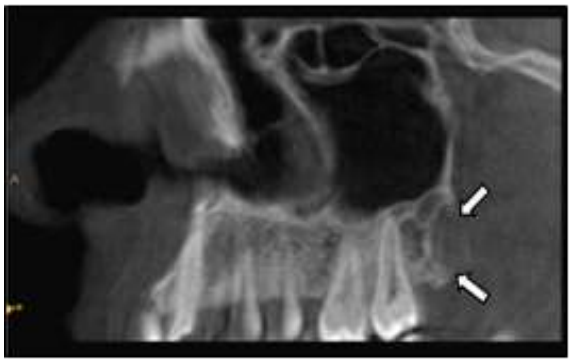
3D View



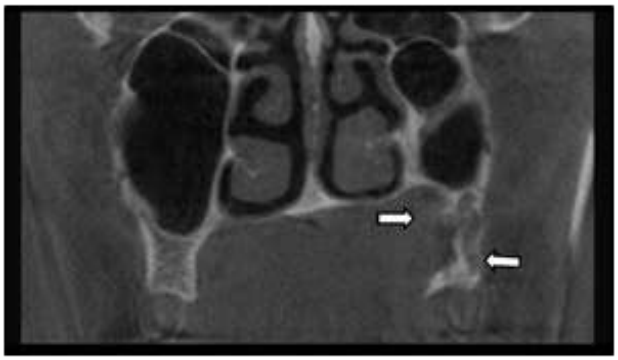
Axial Section



Sagittal section

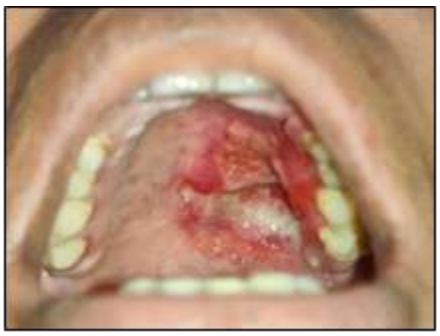


Coronal section



Fine needle aspiration cytology yielded a negative aspiration. An incisional biopsy was performed and histopathological report showed connective tissue stroma with cystic spaces which was lined by varying degrees of cellular proliferation of mucous cells seen. Clear cells containing pale eosinophilic material with centrally places nuclei abundantly throughout the cystic lining suggestive of dysplastic mucous cells. Few cells show hyperchromatic nuclei, cellular and nuclear pleomorphism suggestive of dysplastic epidermoid cells. Overall features were suggestive of low grade mucoepidermoid carcinoma.

Treatment performed was wide local resection of left maxillary region under general anesthesia [Fig. 3].



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[Fig. 3]

Discussion :-

Mucoepidermoid carcinoma (MEC) makes up 5% to 10% of all salivary gland tumors, whereas intraosseous glands comprise only 2% to 4% of all MEC.[1,2,3] There are several current theories explaining the origin of this lesion: (a) ectopic salivary gland tissue

remnants of embryonic salivary glands trapped within the bone; (b) transformation of mucous cells found in odontogenic cysts; and (c) maxillary sinuses or submucosal and mucosal glands with intraosseous extension. Although its etiology is questionable.

Intraosseous mucoepidermoid carcinomas are more common in middle-aged adults and have a slight predilection for females. They are three times more common in the mandible than in the maxilla and are most often found in the area of the molars and mandibular ramus. The most frequently present symptom is cortical bulging, although some lesions may be discovered as an accidental finding on radiographs. Pain, trismus, and paresthesia are

symptoms reported less frequently in these lesions. Metastases have also been reported in 12% of the cases, often as a result of local tumor recurrence, mainly for regional lymph nodes and occasionally for the ipsilateral clavicle, lung, and brain. About 10% of the patients evolve to death[5].

The criteria for central lesions are satisfied by the following findings: radiographic evidence of bone destruction; exclusion of another primary tumour; exclusion of an odontogenic tumour; histopathological confirmation; and detectable intracellular mucins.

Radiographically lesion is described as a radiolucent image with well-defined sclerotic border and numerous small loculations. Also tooth dislocation and root resorption are common findings. Its aggressive behavior is revealed by cortical bone perforation and extension to

surrounding soft tissues. Imaging diagnostic techniques plays very important role in the detection and differentiation of MEC because of its sclerotic periphery and mixed internal structure, consisting of a unilocular and/or multilocular pattern. CT scan is the preferred diagnostic modality as it provides information on the extent and size of the tumor.

Brookestone and Huvos proposed a three-grade system for the classification of intraosseous mucoepidermoid carcinoma[8]:

grade 1- lesions without cortical plate expansion or rupture

grade 2 - lesions with cortical plate expansion but not rupture

grade 3 - lesions with cortical plate rupture and metastasis to nodes

Traditionally, mucoepidermoid carcinomas have been classified into three histopathological grades using the following criteria: quantity of cystic formation, degree of cellular atypia, and relative number of mucous, epidermoid, and intermediate cells.[3]

1) Low-grade tumors exhibit prominent cystic formation, minimal cellular atypia, and a relatively high proportion of mucosal cells.
2) High-grade tumors consist of solid islands of squamous and intermediate cells, which may demonstrate considerable pleomorphism and mitotic activity. Mucus-producing cells may be infrequent, and sometimes, it may be difficult to distinguish the tumor from squamous cell carcinoma.
3) Intermediate grade exhibit characteristics that are located between low and high-grade tumors.[4]

Studies suggest that fine needle aspiration (FNA) is considered to be effective for high-grade or intermediate-grade but unsatisfactory for low-grade. Few cases of calcification have been reported histopathologically. Cases of calcifications in clear cell variants of mucoepidermoid carcinomas and in palatal minor salivary gland mucoepidermoid carcinomas were reported by some authors, and they thought them to be due to dystrophic calcification of the amorphous eosinophilic material secreted by intermediate basal cells. Longstanding cases can manifest with atypical clinical appearances. So, central mucoepidermoid carcinoma should be included in the differential diagnosis of all mixed lesions[4].

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When the correlation between the clinical and histopathological diagnosis was analyzed, only 12.5% of the cases presented a correlation, so the final diagnosis should be based on clinical, radiographic, and histopathological characteristics[7].

Surgery is the main form of treatment. In a review of 64 patients, Brookstone and Huvos observed 40% relapses after conservative surgical modalities such as enucleation, curettage, marsupialization, and marginal resection with or without adjuvant therapy, whereas in the group treated by radical methods such as segmental resection with or without adjuvant treatment associated with the neck, only 4% relapsed[4,5]. Adjuvant therapy, such as radiotherapy and/or chemotherapy, is recommended for high-grade tumors.

Conclusion :-

Although central MEC is a rare neoplasm, we should consider it as one of the differential diagnosis in palatal swellings. It is frequently misdiagnosed, owing to the diverse clinical and radiographic presentations that are indicative of an odontogenic lesion. The tumour is often treated inappropriately or late, eventually leading to recurrence. Advanced imaging greatly aids in timely diagnosis and to identify the involvement of adjacent vital structures, which dictates the management as well as the prognosis. Treated cases of central MEC should be monitored for a longer duration to identify late local recurrence or metastasis.

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