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A rare case of long-standing mucoepidermoid carcinoma of minor salivary glands of the palate

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SUMMARY

Salivary gland carcinomas are the uncommon and clinically diverse group of neoplasms with mucoepidermoid carcinoma (MEC) being the most common among them. MEC accounts for 5% of all salivary gland tumors. As the name implies, the tumor is composed of both mucus secreting cells and epidermoid type cells in varying proportions. Most patients are aware of the lesion for 1 year or less. We report clinical, radiographic and histological features of a long-standing case of mucoepidermoid carcinoma of minor salivary glands of the palate in a 35-year-old male patient. The patient reported with a slow growing swelling on the palate which began 15 years ago. The patient was treated with hemi-maxillectomy and is currently under follow up.

Key words: Mucoepidermoid carcinoma, minor salivary glands, cone beam computed tomography, clear cell.

INTRODUCTION

The malignancies of salivary glands are relatively rare accounting for less than 0.5% of all malignancies and not more than 5% of all head and neck malignancies (1). Mucoepidermoid carcinoma (MEC) is most common malignant salivary gland tumor, first studied and defined as a separate, distinct pathologic entity by Stewart *et al*. in 1945 (2). The tumor is known as "mucoepidermoid carcinoma" because it consists of both the mucous and epidermal cells in different proportions. This tumor shows different biological behaviors, and while the high-grade runs an aggressive course with a poor prognosis, the low-grade type usually demonstrates a more benign nature with an adequate survival rate (3). MEC usually occurs in the age group between third and sixth decades although it can occur at any age and has a slight female predilection. About 53% of MEC occur in major salivary glands; parotid gland is the most common site and accounts for 43% followed by minor salivary glands of palate (4). When MEC arises in minor salivary glands, it can occur on the palate, retro molar area, the floor of the mouth, buccal mucosa, lips, and tongue.

CASE REPORT

A 35-year-old male patient reported to the department of oral medicine and radiology with a chief complaint of swelling of palate since the last 15 years. The patient gave a history of extraction of upper left back teeth around 15 years back. The swelling on the palate followed few months after extraction of teeth and patient immediately consulted a premier dental college and hospital where the patient was admitted for biopsy and was discharged after two days with an impression of a normal histopathological report. Following this, the swelling gradually started to increase in size over the last 15 years, until it reached to the current size. There was no significant pain associated with the swelling but patient complained of nasal discharge since the last 2 to 3 months at the time of reporting to our department. The patient was mentally challenged but there was no dependence on daily activities. The patient at the time of reporting was active and was aware of time, person and surroundings. The patient's family history was non-contributory other than a twin brother who was also mentally challenged. There was no history of trauma, and the patient was under medication after undergoing a psychiatric evaluation a few years back.

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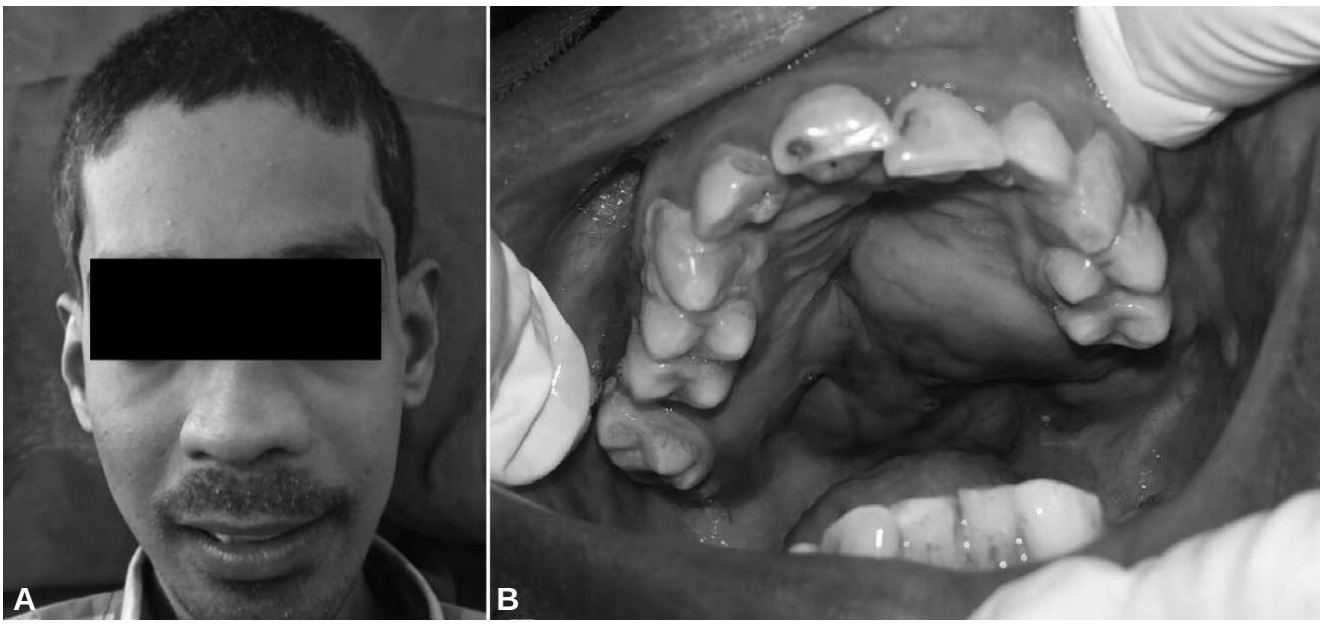

Fig 1. A – swelling seen on upper lip with deviation of nasal bridge towards the left; B – palatal swelling crossing the junction of hard and soft palate.

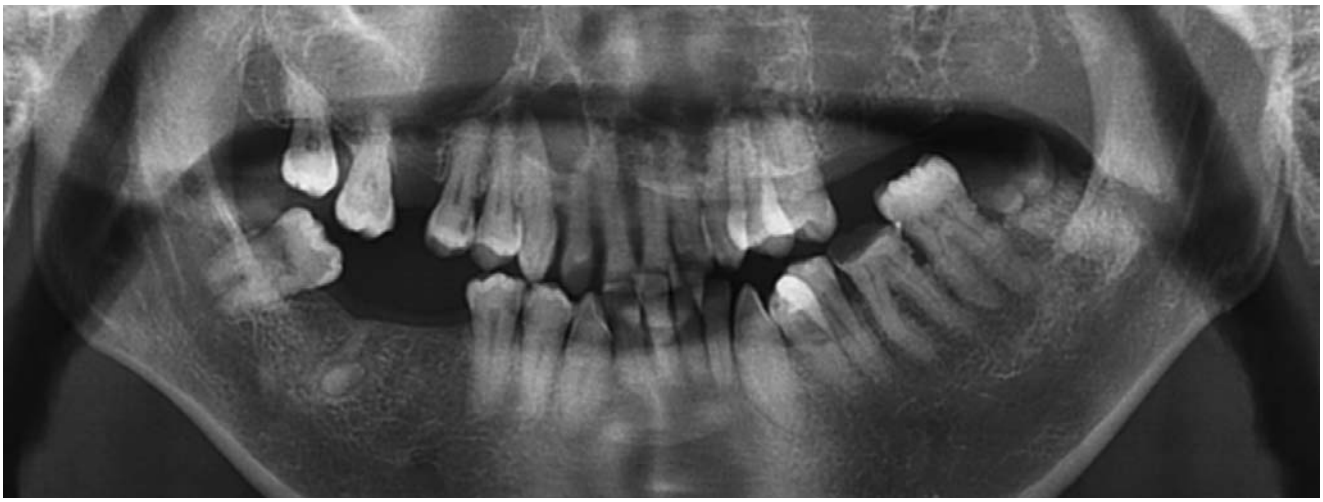

Fig 2. Panoramic radiograph revealing ill defined multilocular lesion in maxilla with complete involvement of left maxillary sinus and lesion crossing the midline towards the right

On clinical examination, the patient had a diffuse extra-oral swelling over upper left half of lip measuring around 3×3 cm in dimensions and patients nasal bridge could be seen deviated towards the right side (Figure 1, A). On intraoral examination, there was a bony hard swelling over the labial cortical plate with respect to teeth number 21, 22, 23 and 24 and a diffuse swelling around 5×4 cm over Left half of palate (Figure 1, B). The palatal swelling extended anteriorly from 1.5 cm posterior to palatal aspect of teeth number 11 and 12 and crossed the junction of hard and soft palate posteriorly. Mediolaterally the swelling extended from the midline of the palate and extended towards the left side crossing the edentulous ridge in the region corresponding to the position of teeth number 26 and 27. The swelling was pinkish anteriorly, while it was purple in color posteriorly. There were no secondary changes noted, and it was non-tender on palpation. The swelling was soft to firm in consistency. Benign tumor of minor salivary glands of the palate was put as a provisional diagnosis.

Panoramic radiograph showed ill-defined multilocular lesion in maxilla with trapped irregular trabeculae, and destruction of the medial wall and floor of the left maxillary sinus (Figure 2). The lesion can be seen involving the right maxillary sinus. As the lesion seemed to be quite a destructive, cone beam computed tomography (CBCT) was utilized to understand the extension of the lesion. After taking consent for CBCT exposure, image acquisition was done using CBCT Planmeca Promax 3D Mid (Planmeca Finland) machine at 90 kVp,8 mA, 13.772 seconds scan time, 20.2 cm

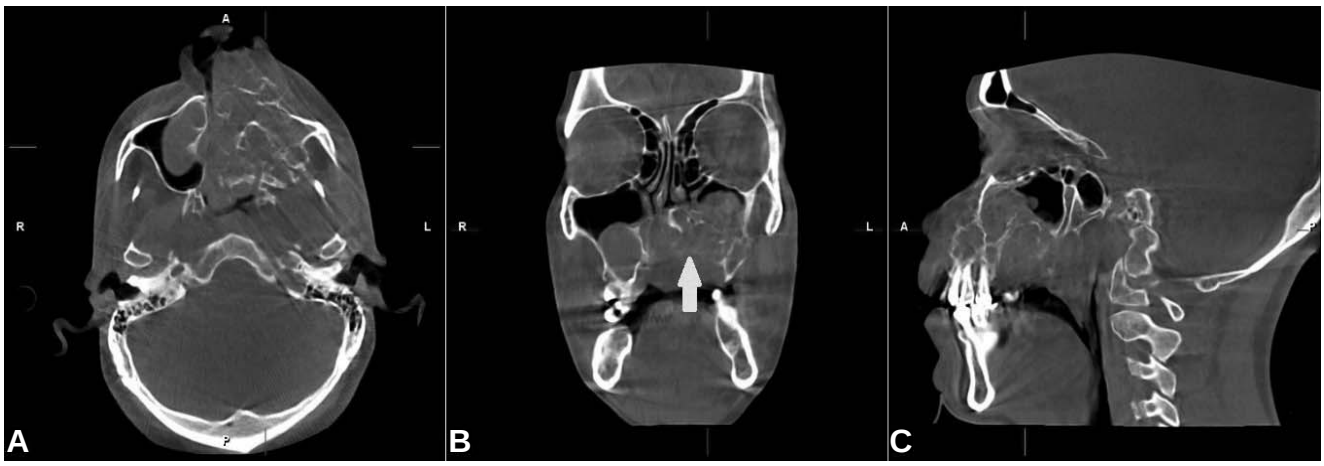

Fig 3. A – CBCT axial section showing multilocular lesion with crenated borders extending and involving the left pterygoid plates; B – coronal section showing destruction of palate (Arrows); C – sagittal section shows undermining and infiltrative growth of multilocular lesion.

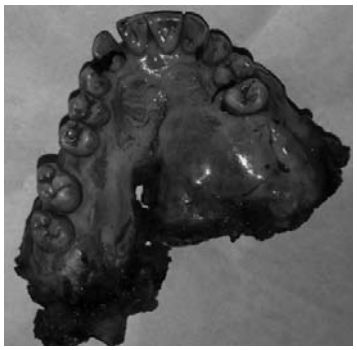

Fig 4. Custom abutment – model view

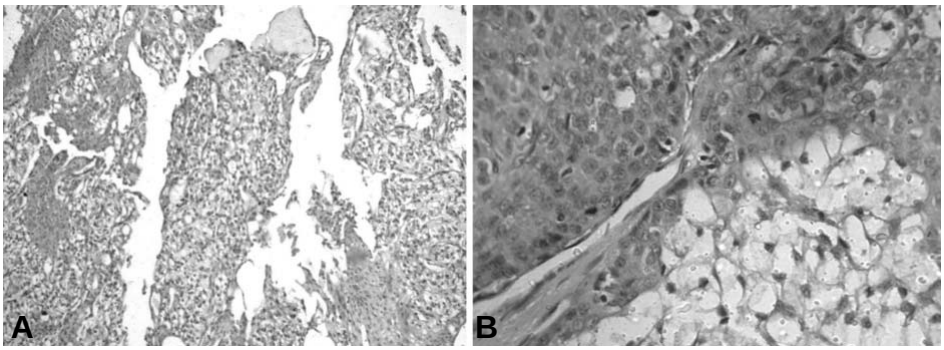

Fig 5. A – H&E ×100 shows cyst like areas, epidermoid, intermediate and clear cells; B – H&E ×400 shows large polygonal cells with clear cytoplasm and sharply defined cytoplasmic borders along with centrally placed nuclei.

height and 17.5 cm diameter scan volume. CBCT axial section showed an expansile multilocular pattern with irregular borders involving right and left maxillary sinus, with the destruction of anterior, medial and lateral walls of the left maxillary sinus with its complete obliteration (Figure 3, A). The lesion can be seen crossing the midline and involving the right maxillary sinus with a deviation of nasal septum towards the right side. Coronal section of CBCT shows the destruction of the palate (Arrow) and multilocular lesion within left maxillary sinus involving its medial wall, lateral wall and the floor (Figure 3, B). The lesion in this section can be seen crossing to the right side with the involvement of nasal cavity, medial wall of right maxillary sinus and its opacification. The sagittal section shows multilocular mass having thick and thin internal cortical septae resembling a honey comb in addition to the destruction of the superior crest of the alveolar process (Figure 3, B). Based on radiological features, we put a radiographic diagnosis of Ameloblastoma.

Incisional biopsy was carried out and hematoxylin and eosin (H & E) stained section revealed a large number of cystic spaces filled with mucin, in addition to large sheets of epidermoid and intermediate cells. Epidermoid cells also showed clear cell differentiation. All these features suggested of epidermoid carcinoma. Hemi-maxillectomy was carried out, and an obturator was fabricated to rehabilitate the surgical defect. The resected specimen (Figure 4) was sent for histopathological investigations which revealed cyst like areas, epidermoid, intermediate and clear cells when stained with hematoxylin and eosin under a magnification of 100 (Figure 5, A). The biopsy specimen upon staining with haematoxylin and eosin and under a magnification of 400 showed cystic spaces filled with eosinophilic material, surrounded by intermediate cells, large islands of epidermoid cells with sheets of large polygonal cells having clear cytoplasm and sharply defined cytoplasmic borders along with centrally placed nuclei. (Figure 5, B). Mucoepidermoid carcinoma of clear cell variant was put as the final diagnosis based on the histological investigation of the resected specimen, and this variant of MEC is considered as a rare presentation of MEC. The patient was evaluated every week for the initial period

of 2 months of surgery and showed good signs of recovery. The patient is currently under follow-up.

DISCUSSION

Mucoepidermoid carcinoma (MEC) is known to have an uncertain pathogenesis, although several underlying etiologies have been suggested including prior radiation exposure, to reciprocal translocation of t (11;19) that leads to the formation of CRTC1-MAML2 fusion oncogene (5). MEC of salivary glands is reported to initiate from pluripotent reserve cells of salivary gland ductal excretory system that can differentiate into mucous, columnar, and squamous cells (6). It has been seen that few of previously benign tumors of epidermoid origin subsequently metastasize and hence all such benign tumors are considered as carcinoma. MEC was first described by Massao and Berger in 1942 (7) and was recognized as a separate entity among salivary gland neoplasms by Stewart et al. in 1945. MEC occurs mostly between 50 to 60 years of age, in parotid gland and palate on a majority of instances when affecting major and minor salivary glands respectively (8). The histological grade dictates the clinical presentation of MEC. The lesions of low and intermediate grade grow slowly, while as the lesions having high histological grade are rapidly enlarging (9). MEC is asymptomatic in a majority of the cases with clinical manifestation of an asymptomatic swelling with only a few cases being symptomatic. The patient in this case report was largely asymptomatic with a gradual increase of palatal swelling over the period of last 15 years.

The study of MEC can be done by variously advanced imaging modalities including Magnetic Resonance Imaging (MRI), Multi Detector Computed Tomography (MDCT), and Cone Beam Computed Tomography (CBCT), and lesions extension and any bone invasion can be assessed using CBCT or CT scans (10). CBCT has an advantage over MDCT when it comes to radiation dose and cost (11). Cross-sectional images with MDCT or CBCT in cases suspected of malignancies should be attained; furthermore, it has been seen that images acquired by CBCT are as reliable as MDCT in predicting the invasion of bone in malignant lesions (12). All the above-mentioned advantages of CBCT were utilized in the present case and the multiplanar CBCT images showed the extension of the lesion and the bony invasion.

MEC has no typical histopathological features and is composed of epidermoid and mucin producing cells originating from the epithelial lining of ducts of salivary glands. Histologically, the tumor resembles squamous cell carcinoma if epidermoid cells predominate and the variant is thus called as high-grade MEC, where as in case of more of mucin producing cells, with a preponderance of cystic architecture the variant is labeled as a low-grade MEC. Clear cells are inconsistently sized and shaped having sharp outlines and a hydropic, water-clear cytoplasm with their nuclei being small, vesicular or pyknotic, in a central location, furthermore occasionally, it seems that focally, there is a transition between epidermoid and clear cells (13). MEC of clear cell variety makes its distinction from other clear cell tumors a challenging task. This case showed positive staining with a special stain of mucicarmine and Periodic Acid Schiff stain.

The differential diagnosis for MEC includes, but is not restricted to, necrotizing sialometaplasia, adeno- squamous carcinoma, cystadenoma, cystadenocarcinoma, sebaceous carcinoma, inverted papilloma, squamous cell carcinoma (high-grade tumors) and other clear cell tumors such as hyalinizing clear cell carcinoma, acinic cell carcinoma, metastatic renal cell carcinoma and clear cell oncocytoma, (14).

The traditional method of treatment for a majority of MEC involving the palate has been the surgical management of palatal minor salivary gland tumors, whereas wide local excision and enucleation has been the most widely followed procedure for benign and malignant lesions (6). It is recommended to resect malignant MEC completely in one attempt, with an adequate margin of normal tissue, considering the higher rates of recurrences as quoted in literature in the lesions with positive margins or in lesions declared positive initially, that turned out to be negative eventually (15). As our case had extensive bone involvement, we had to resort to hemi-maxillectomy with pedicle flap taken from temporalis, to cover the surgical defect followed by obturator placement. Recurrences if any occur mostly in the first year of treatment and the survival rate being 92% for the low grade, 83% of the intermediate grade and 24% for high-grade tumor (16). The patient in this case report is currently under follow up.

CONCLUSION

We reported a long-standing case of mucoepidermoid carcinoma of the palate. CBCT gave us an idea about the extensions of the lesion and together with histopathological report helped us to devise an

appropriate surgical procedure which seems to be a success as the patient is free of any recurrences after 8 months of follow-up.

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Conflict of interest: None declared.

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