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CASE REPORT

Mucoepidermoid carcinoma involving the palate with lamellated calcifications: A notable finding

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ABSTRACT

Mucoepidermoid carcinoma (MEC) is a malignant glandular epithelial neoplasm of the salivary glands with an unpredictable behavior and a tendency to recur. The tumor is composed of mucous secreting cells, epidermoid cells, intermediate cells, columnar cells, and clear cells in varying proportions. Calcification in the salivary gland tumors is less common and minuscule in size; these can be seen in the benign tumor-like pleomorphic adenoma to malignant tumor-like MEC. Calcifications in MEC are considerably sparse and commonly associated with high-grade tumors. We present a case of MEC with lamellated calcifications present in the palate of a 43-year-old male patient.

Key words
Calcifications, mucoepidermoid carcinoma, salivary glands

INTRODUCTION

Mucoepidermoid carcinoma (MEC) is the most common malignant salivary gland tumor. It accounts for 5–9% of all salivary gland neoplasms.[1] It has a distinct female predilection of 3:2. Approximately half of the tumors occur in the major salivary glands; commonly seen in the parotid gland and minor salivary glands of the palate.[2] Central MEC's of the mandible are also reported. It is a very slowly growing tumor, and patients are aware of its presence only after several years of its onset. MEC arise from the reserve cells of the salivary duct system. Therefore, they have the propensity to differentiate into mucin-producing cells or duct-like epidermoid cells, both of which are altered neoplastic cells. MEC can be graded as low-, intermediate-, and high-grade varieties based on the existence of the predominating cell population of mucous cells, intermediate cells, and epidermoid cells.[1]

When the soft-tissue calcification occurs with elevated serum levels of calcium-phosphorus ions, it is termed metastatic calcification. When soft-tissue calcification occurs in altered, necrotic, or dead tissue with normal serum calcium and phosphorus levels, it is called dystrophic calcification.[3]

Calcification in the MEC has been considered to be dystrophic calcification. Due to the lack of data, it is accredited that calcifications in MEC would be associated with high-grade tumors giving in aggressive outcomes.[4] This article reported a case of MEC with lamellated calcifications and discussed their possible mechanism of formation.

CASE REPORT

A 43-year-old male patient reported to the outpatient department with a chief complaint of an ulcerated swelling in the right palatal region. The patient was aware of the presence of lesion since 10 years. The lesion was

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not associated with pain and discharge. The patient had a habit of smoking seven cigarettes per day for the last 20 years. Past medical history revealed that the patient was hypertensive, a hyperthyroid, and presently was under medication for the above and also for gastritis.

Intraoral examination revealed a well-defined erythematous swelling with surface ulceration on the right half of the palate. The swelling was roughly oval in shape measuring approximately 3 cm × 2 cm in size. Anteroposteriorly, the lesion was located 1.5 cm away from the incisors and extended up to the junction between the soft and hard palate. Mediolaterally, it extended from the mid-palatine raphe up to 0.5 cm from the palatal gingival margin [Figure 1]. On palpation, the swelling was non-fluctuant, soft to firm in consistency, tender, movable, and not fixed to the underlying structure. Based on these features, a differential diagnosis of a malignant salivary gland tumor such as adenoid cystic carcinoma, MEC, or squamous cell carcinoma of the palate was given.

Routine blood investigations along with occlusal and orthopantomograph radiographs were advised, which were not significant. Computed tomography scan of maxilla and neck revealed faint enhancing lesion of 26 mm × 11 mm seen in the soft tissues of the hard palate at the right of the midline. Thinning of the underlying hard palate and a few small calcific densities within the mass were observed [Figure 2]. Focal erosion of the right hard palate associated with 1.9 cm × 1.2 cm × 1.6 cm nodular enhancing soft tissue along the oral surface was seen. Few focal nodular calcifications were also reported.

Histopathological examination showed sheets and islands of proliferating glandular epithelial cells in the connective tissue stroma underneath a parakeratinized stratified squamous epithelium. The glandular epithelium showed a wide range of mucous, epidermoid cells with areas of cystic degeneration. Intermediate cells were seen occasionally. Numerous hematoxyphilic, concentrically lamellated calcifications analogous to psammoma bodies were evident within the lesional tissue [Figure 3a and b]. Based on the histopathological features, it was diagnosed as MEC -Low grade.

Right partial maxillectomy was done, and the patient was followed up every 15 days for 3 months and later every month for 1 year during which he remained asymptomatic.

Figure 1: Photograph showing ulcerative growth extending up to junction between soft and hard palate

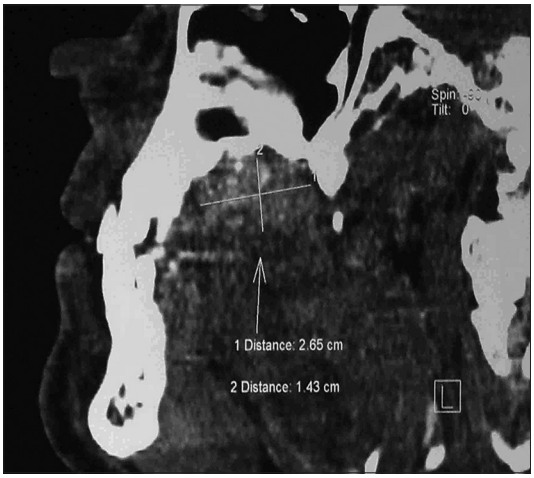

Figure 2: Enhanced computed tomography scan of the maxilla and neck showing thinning of the underlying hard palate and a few small calcific densities within the mass

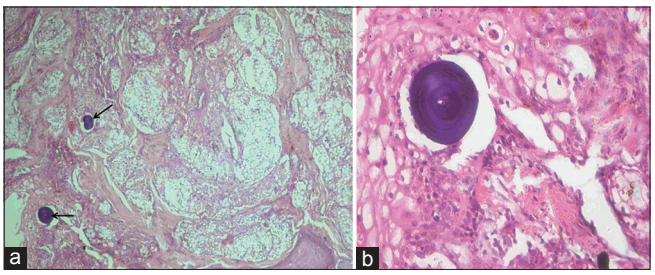

Figure 3: (a) Photomicrograph showing hematoxyphilic concentrically lamellated calcifications under ×10. (b) Photomicrograph showing hematoxyphilic concentrically lamellated calcifications under ×40

DISCUSSION

Stewart *et al.* described MEC as a mucoepidermoid tumor that includes a "benign tumor" and "malignant tumor" with metastatic potential, which now represent low and high grade, respectively. Later, the intermediate grade was included in the grading system.[5] MEC is the most common salivary gland malignancy in both children and adults. About 70% of these tumors are reported in the parotid gland and 6–10% in the submandibular gland and remaining in the minor salivary glands of the palate. MEC constitutes 26% of all malignancies of the palate. MEC is the most common intraosseous salivary gland tumor which is usually asymptomatic; it has a female predilection, more commonly seen in

the mandible than in the maxilla in the region of the third molar. It is believed that these tumors arise from the odontogenic cysts or entrapped salivary gland tissue during embryological development.[6] Based on histological grades, MEC comprises different proportions of mucous, epidermoid, and intermittent cells and has abundant pale, foamy cytoplasm that shows positivity for mucin stains (mucicarmine and periodic acid-Schiff). The epidermoid cells have a polygonal shape with ovoid to elongated or vesicular nuclei with abundant eosinophilic cytoplasm and intercellular bridges. Occasionally, they are associated with keratin pearls. The intermediate cells are highly prolific, basaloid cells which are larger than the basal cells and smaller than the squamous cells and are thought to be the progenitor of epidermoid and mucous cells.[7]

The present case showed classical histopathology of MEC comprised sheets and islands of proliferating glandular epithelial cells in the connective tissue stroma. A notable finding in the case was the presence of a number of hematoxyphilic concentrically lamellated calcifications within the lesional tissue. Very few cases of MEC with calcification are reported in literature. It is noticeable to mention that all of the previous tumors reported in literature affected minor salivary glands, preponderance cases involved the minor salivary glands of the palate.[4] In the present case, calcifications were observed in MEC of minor salivary gland of the palate, which is in accordance with literature.

Although calcification is a common finding in inflammatory salivary gland disorders, salivary glands tumors rarely show calcifications. González-Arriagada *et al.*[4] examined thirty cases of MEC for calcifications and found only six cases with calcification, of which five cases were conventional and one clear cell MEC while most of the cases were present in the minor salivary glands. He suggested that calcification is because of dystrophic calcification of mucin secreted by the malignant cells. Yoon *et al.*[8] proposed four mechanisms to explain the calcification process in MEC. They may form as a result of hypercalcemia, dystrophic calcification of necrotic areas as a component of the tumor, or by the calcification of the material secreted by tumor cells. Siar *et al.*[9] reported an unusual case of MEC showing calcifications and suggested that these structures are laid down in layers and probably represent some form of dystrophic calcification.

Five cases of MEC with calcification were found in the minor salivary glands of the oral cavity. This observation may suggest that calcifications are more likely to develop in tumors affecting the mucous salivary glands. As the palatine salivary glands are pure mucous in nature, it emphasizes the theory that calcifications may originate from the precipitation of salivary gland mucous secretion.[4] The significance of calcifications is not clear. According to literature, calcifications are associated with high-grade tumors with aggressive outcome. A few studies have concluded that calcifications are not related to histopathological grade of differentiation and not associated with outcome.[10]

MEC on palate should be differentiated from all benign and malignant tumors of the hard palate, mostly pleomorphic adenoma, polymorphous low-grade adenocarcinoma, adenoid cystic carcinoma, and squamous cell carcinoma. Chronic sialadenitis or mucocele, which is histologically similar, could be misdiagnosed as a low-grade MEC. Odontogenic cysts, lymphoma, plasmacytoma, Langerhans cell histiocytosis, or metastatic carcinoma, as well as a rare papillary oncocytic cystadenoma, could also be included in the differential diagnosis. The prognosis is dependent on the clinical stage, site, grading, and adequacy of surgery.[1]

The overall recurrence rate of MEC is about 25%, Thorvaldsson *et al.*,[11] have reported that the recurrence rate is 10% for low-grade lesions but 74% for high-grade tumors.

CONCLUSION

Salivary gland tumors rarely show calcifications in the parenchymal tissue; however, it is commonly seen in the inflammatory disorders of the salivary gland. The calcification seen in the case could be dystrophic in nature, formed from the mucin secreted by the malignant cells. Occurrence of such calcification in only sporadic salivary gland tumor leaves us denuded of perpetual focus on their influence on prognosis and recurrence, if any.

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Conflicts of interest
There are no conflicts of interest.

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