Case Report

Predominantly oncocytic mucoepidermoid carcinoma of palate: A case report

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Abstract

Oncocytic mucoepidermoid carcinoma (OMEC) is an uncommon variant of mucoepidermoid carcinoma. Histopathologically, it is characterised by the predominance of cells with large polygonal morphology and with an abundance of eosinophilic granules. We present a rare case of OMEC manifested as painless palatal swelling in a 25-year-old young male. The overlying mucosa was normal in appearance, with no evidence of ulceration or discharge. Histopathology examination showed the presence of sheets of mucous and intermediate cells along with cystic areas of variable sizes and shapes. On high power magnification, oncocytes were evident, showing abundant granular eosinophilic cytoplasm with central dark round nuclei. Around 75–80% tumour cell population was composed of oncocytic cells. The predominant presence of oncocytes can present diagnostic difficulties to pathologists due to overlapping features with adenoid cystic carcinoma, oncocytoma, acinic cell carcinoma, Warthin's tumour, and other oncocyte tumours. Although the presence of oncocytes is a pathognomonic feature, the role of immunohistochemistry and genetic analysis in diagnosis is discussed in the present paper. Moreover, considering its behaviour as a low-grade MEC, it is prudent to avoid an aggressive treatment strategy and prevent unwarranted morbidity. We recommend prospective studies to better understand the factors that influence the prognosis of OMEC.

Keywords: MAML2 rearrangement, oncocytic mucoepidermoid carcinoma, palatal swelling

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INTRODUCTION

Salivary gland tumours are heterogeneous in nature, yet cause for concern due to their proximity to the orofacial structure. Further, these tumours show diverse clinical courses and biological behaviour based on their histopathology. These tumours are themselves rare in the oral cavity. However, when it occurs, it poses a diagnostic dilemma to the clinician and oral pathologist. Irrespective of benign or malignant nature, they share a common origin, resulting in overlapping histomorphology, further leading to unpredictability in tumour behaviour, especially in mucoepidermoid carcinoma (MEC).[1,2]

MEC is histologically characterised by the presence of different cell types, such as mucous, squamous, intermediate, clear, and epidermoid, with varied arrangements surrounding the cystic spaces.[3] This diversified and

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heterogeneous cellular morphology represents the plasticity in the tumour cells. However, oncocytic differentiation in MEC is considered an uncommon phenomenon.[4] Since oncocytes are a feature of other types of salivary gland tumours, sometimes differential diagnosis could be problematic. Further, it suggested that it might influence the prognosis and course of the disease. Unfortunately, being a rare entity, little evidence is available on oncocytic MEC (OMEC) and its biological behaviour. In the present paper, a case of OMEC in a young male patient with palatal swelling is presented.

CASE REPORT

A 25-year-old male patient reported with a chief complaint of painless, slow swelling on the right side of the palate for one and a half months. The swelling was 1 × 1.2 cm^2 in size and soft to firm consistency [Figure 1a]. It extended anteroposteriorly from the premolar region to the third molar region and mediolaterally from the gingival margin to the mid-palatine raphe. The overlying mucosa was normal in appearance, with no evidence of ulceration or any discharge. The patient's medical and family history was unremarkable.

The microscopy examination on the incisional biopsy showed an unencapsulated tumour with numerous cystic and solid areas. The lesional tissue showed the presence of sheets of mucous and intermediate cells along with cystic areas of variable sizes and shapes [Figure 1b]. The majority of the cystic spaces were lined by frank oncocytic cells. On high power magnification, oncocytes showed abundant granular eosinophilic cytoplasm with central dark round nuclei [Figure 1c and d]. Around 75–80% tumour cell population was composed of oncocytic cells. Fibrous stroma and inflammatory infiltrate were present at the periphery of the tumour. Due to the predominant oncocytic cell presence, the lesion was diagnosed as OMEC. The lesion was surgically excised under general anaesthesia, and the features of the excised specimen were consistent with the findings of incisional biopsy. There was no evidence of recurrence after 1-year follow-up of the patient.

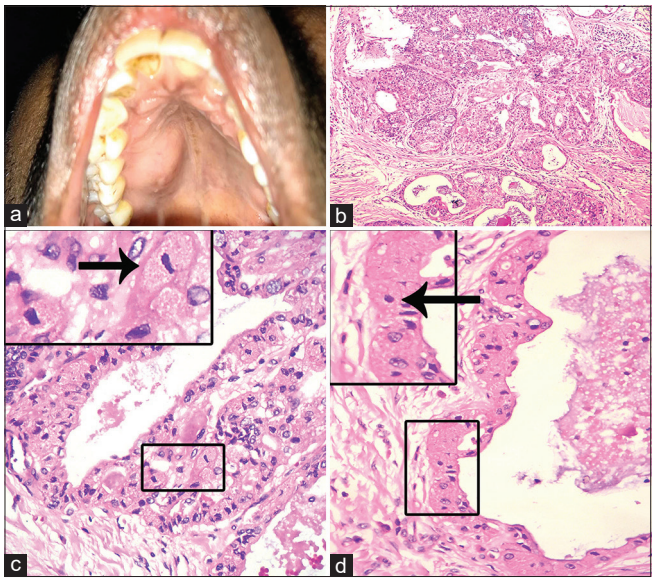

Figure 1: (a) Intraoral preoperative clinical picture showing swelling involving the right side of the hard palate with normal overlying mucosa. (b) Unencapsulated tumour showing sheets of mucous and intermediate cells along with cystic areas of variable sizes and shapes (hematoxylin-eosin, Total magnification x100). (c and d) The cystic spaces were lined by oncocytic cells showing abundant granular eosinophilic cytoplasm with central dark round nuclei (black arrow). Inset in c and d shows a higher magnified view of the cells from the select areas. (hematoxylin-eosin; total magnification x400)

DISCUSSION

MEC has the potential for aggressive destruction, although it has better survival and prognosis depending upon the site and histological grade or features.[5] Low-grade MEC has a better prognosis as compared to high-grade MEC.[6] Low-grade MEC contains a more cystic component with mucous secreting cells, whereas high-grade has solid components and pleomorphic cells. However, sometimes, the presence of non-pathognomonic cells such as oncocytes may be seen as observed in the present case. In literature, the majority of OMEC cases were reported in the parotid gland; however, cases of palate OMEC are also documented.[7] OMEC of the palate is slow-growing and can be asymptomatic for a few years. It can occur between the 1st decade and the 8th decade. A slight male preponderance was reported in the literature.

DIAGNOSTIC CHALLENGE

These oncocytes proliferate and predominate as compared to other cells in MEC, giving a resemblance to oncocytic tumours.[8] Further, these features overlap with adenoid cystic carcinoma, oncocytoma, acinic cell carcinoma, Warthin's tumour, and epithelial-myoepithelial carcinoma, posing difficulty in diagnosis.[9] However, the presence of characteristic cystic areas and mucous cell population in some areas leads to the definitive diagnosis. In contrast, tumours such as adenoid cystic carcinoma show a typical cribriform or tubular pattern of cells with abundant clear or eosinophilic cytoplasm. Oncocytoma is characterised by a solid tumour composed of oncocytes that are usually arranged in acini or cords. In contrast, acinic cell carcinoma shows a solid or cystic tumour composed of acinar cells with abundant granular cytoplasm. Oncocytes, along with

Table 1: Key diagnostic features of oncocytic mucoepidermoid carcinoma and other oncocytic tumours

Tumour type	Key diagnostic features	IHC markers
Oncocytic mucoepidermoid carcinoma	Mixture of mucus cells, intermediate cells, squamoid or epidermoid cells, and predominant oncocytes.	Positive for p63, EMA, and S100. Negative for CK7 and CK20.
Adenoid cystic carcinoma	Cribriform or tubular pattern of cells with abundant clear or eosinophilic cytoplasm.	Positive for p63, EMA, and S100. Negative for CK7 and CK20.
Oncocytoma	composed predominantly of oncocytes. The oncocytes are usually arranged in islands or cords. Other cell types are absent.	Positive for p63, EMA, and S100. Negative for CK7 and CK20.
Warthin's tumor	Composed of oncocytes and lymphoid tissue. The oncocytes are usually present in the lining epithelium. There is usually a prominent lymphoid component.	Positive for p63, EMA, and S100. Positive for CD20, CD3, and PAX5. Negative for CK7 and CK20.
Clear cell oncocytoma	Clear oncocytes are usually arranged in acini or cords. Occasionally, there could be a presence of granular cytoplasm as a diagnostic feature.	Positive for p63, EMA, and S100. Negative for CK7 and CK20.
Epithelial-myoepithelial carcinoma	Tumors composed of oncocytes, myoepithelial cells, and other cell types. The oncocytes are usually arranged in acini or cords. There is usually a prominent myoepithelial component.	Positive for p63, EMA, and S100. Positive for calponin and smooth muscle actin. Negative for CK7 and CK20.

CK7=Cytokeratin 7, CK20=Cytokeratin 20, EMA=Epithelial membrane antigen, IHC=Immunohistochemistry, PAX5=Paired box 5

lymphoid tissue arranged in acini or cords, are features of Warthin's tumour [Table 1].

The role of immunohistochemistry in the diagnosis of OMEC is non-conclusive, although a few authors suggested that p63 might aid in differentiating it from other oncocytic lesions. p63 shows diffuse and strong positivity. However, the presence of mastermind-like 2 (MAML2) rearrangement can distinguish OMEC from other oncocytic lesions.[10] Since these markers might not be available in day-to-day practice, and hence careful identification of the characteristic features of MEC is the key to the diagnosis. To further simplify the diagnostic approach, the key histopathology and immunohistochemistry (IHC) features of OMEC and other oncocytic tumours are explained in Table 1.

Prognosis

Unfortunately, the impact of oncocytes on the progression of lesions is mostly unknown. Weinreb *et al*.[11] observed that OMEC behaviour is similar to low-grade MEC. Hence, local surgical excision can be sufficient for a better outcome in terms of recurrence.[7] The overall 5-year survival rate for OMEC is 90%. However, the prognosis can vary depending on the tumour grade, stage, and extent of surgery. Patients with high-grade OMEC or those who have tumours that have spread to lymph nodes or other parts of the body have a worse prognosis.[11] Liao *et al*.[12] reported on a case of OMEC that was positive for the MAML2 gene rearrangement. This gene rearrangement is associated with a more aggressive tumour behaviour, but the patient in this study had a good prognosis with no evidence of recurrence after 5 years.

Future Directions

We recommend investigating the molecular mechanisms that drive OMEC development and progression. This could lead to the development of new targeted therapies for OMEC. Moreover, prospective studies are recommended to better understand the factors that influence the prognosis of OMEC. This could help to identify patients who are at risk for recurrence or metastasis. OMEC is a rare cancer, so research in this area is limited. However, there is growing interest in OMEC, and these future directions will lead to significant advances in our understanding and treatment of this disease.

CONCLUSION

This case report summarises the distinguishing characteristics of OMEC and the diagnostic difficulties it presents, notably because of the presence of oncocytes, which can mimic other oncocytic salivary gland tumours. It emphasises the value of using IHC and genetic analysis in the diagnostic procedure, underlining the demand for a thorough and accurate method to identify OMEC. In addition, knowing that it is a low-grade mucoepidermoid carcinoma motivates a cautious therapeutic approach to avoid needless morbidity and highlights the importance of individualised treatment regimens for uncommon malignancies like OMEC. In order to guarantee an accurate diagnosis and suitable clinical therapy, it is crucial to grasp the distinctive features of OMEC, as demonstrated by this case.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest
There are no conflicts of interest.

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