



**ORIGINAL RESEARCH PAPER**

**ENT**

**A RARE CASE REPORT OF MUCOEPIDERMOID CARCINOMA – RETROMOLAR REGION**

**KEY WORDS:** Salivary gland tumours Mucoepidermoid Carcinoma Retromolar trigone Histological grade

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**ABSTRACT**

Salivary gland tumours constitute 5% of head and neck cancers of which Mucoepidermoid Carcinoma (MEC) is the most common, representing 10% of all salivary gland tumours. The subsites of MEC in oral cavity are the palate, buccal mucosa, retromolar trigone and upper lip. Its presentation in the retromolar region is a rare entity with about 6.4%. It has diverse biological characteristics, correlated to the histological grade of the tumour, graded as low, intermediate and high grade. It has good outcome after surgical management, with a prognosis related to the histological grade and stage of the tumour. Low-grade subgroup without further negative prognostic features can be managed with surgery alone. The intermediate-grade group patients will have additional negative prognostic factors and is an indication for adjuvant radiotherapy. Here we report a case of MEC, in the retromolar region of low grade treated surgically without any recurrence in the follow up.

**INTRODUCTION**

Salivary malignancies account for around 5% of malignancies of the head and neck. Mucoepidermoid carcinoma (MEC) is the most common salivary malignancy in most series, accounting for roughly 45% of cases. Between 50 and 70% arise in the parotid gland, 15–35% in oral cavity minor salivary glands and 6–11% in the submandibular glands. Patients of all ages and both sexes are affected.<sup>5</sup> The most common subsites in oral cavity are the palate, buccal mucosa, retromolar trigone and upper lip, accounting for ± 75% of cases. The palate is the most common site (55%) with ± 60% of these being malignant.<sup>6</sup> The localization at the retromolar trigone represents 6.4% of minor salivary gland tumours.<sup>2</sup> We report a case of MEC in an adult female presenting at retromolar region.

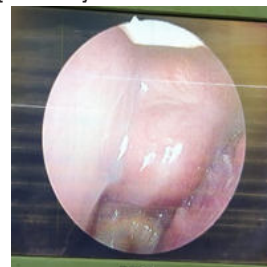
**Case report**

A 33 years old female patient presented to us with swelling in the right retromolar region for 2 years. The swelling was gradually increasing in size with time associated with mild pain occasionally. Clinical examination revealed a solitary smooth oval swelling of 3.5 x 2.5 cm in the right retromolar trigone region, with well-defined borders and firm consistency, not mobile and not tender, and with an erythematous mucosa (Fig.1). Regional lymph nodes were not palpable.

FNAC of the swelling was done which showed epithelial cells in clusters with macrophages and inflammatory cells against mucin background. CECT neck showed ill-defined mixed solid cystic lesion with enhancing solid area in right retromolar region measuring 3 x 2.1 x 1.1 cm suggestive of inflammatory/ neoplastic lesion. No bony erosions noted (Fig.2). MRI neck was taken which showed a well-defined lobulated solid cystic lesion in right retromolar region measuring 3.1 x 2.4 x 2.6 cm with small T2 hyperintense cystic areas and small T2 hypointense internal solid areas, suggestive of minor salivary gland tumour (Fig.3).

With the provisional diagnosis of minor salivary gland tumour in retromolar region, patient was planned for surgery under general anaesthesia after all routine blood investigations. Wide local excision of the swelling with 1.5 cm margin was performed (Fig.4) and the specimen was sent for histopathological examination. Postoperative period was uneventful. HPE showed extensive areas of hyalinized fibrous tissue with scattered glands lined by mucinous epithelium and intermediate type cells with focal areas of cholesterol clefts with lymphoid aggregates, with features suggestive of MEC of minor salivary gland with margins of the specimen clear of tumour cells (Fig.5). The patient was asked to follow

up monthly for 6 months and we found no recurrence.



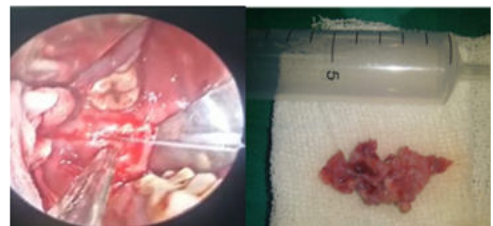
**Fig.1: Swelling in right retromolar region**



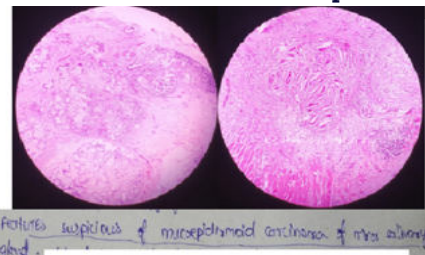
**Fig.2: CECT neck and PNS**



**Fig.3: MRI Head and Neck**



**Fig.4: Wide local excision with resected specimen**



**Fig.5: HPE of the specimen showing intermediate cells,**

**mucous cell and epidermoid cells with surrounding lymphoid aggregates**

**DISCUSSION**

The tumours of the retromolar trigone region represent 12% of neoplasms of the oral cavity. They are represented by squamous cell carcinoma in the majority of cases (>95%), adenocarcinoma, adenoid cystic carcinoma, and mucoepidermoid carcinoma which is rare.<sup>2</sup>

Mucoepidermoid carcinoma (MEC) is the most common salivary malignancy in most series, accounting for roughly 45 per cent of cases. Between 50 and 70% arise in the parotid gland, 15–35% in oral cavity minor salivary glands and 6–11% in the submandibular glands.<sup>5</sup> The most common subsites in oral cavity are the palate, buccal mucosa, retromolar trigone and upper lip, accounting for ± 75% of cases. The palate is the most common site (55%) with ± 60% of these being malignant.<sup>6</sup> The localization at the retro molar trigone represents 6.4% of minor salivary gland tumours.<sup>2</sup> The mean age is approximately 45 years, with a slight predilection for women, in a 3:2 female/male ratio. Nevertheless, this women predominance is more pronounced, mainly, in tumours of the tongue and retromolar area.<sup>4</sup> The environmental and nutritional factors, CMV infection have been linked to the development of MEC.<sup>6</sup>

The tumorigenesis of MEC is said to follow 'Reverse cell theory', arising from the excretory duct reverse cell, due to problem in normal differentiation process.<sup>5</sup> The clinical behaviour of MEC is widely variable, ranging from indolent tumour growth to highly aggressive metastatic dissemination. Many MEC lesion presents as small solid mass or soft tissue lesion with granular or papillary surfaces and ulcerated lesions. Some may present as bluish or red-purple, fluctuant, smooth surfaced mass, which appear very similar to mucocele. They are generally slowly progressive asymptomatic lesions with history lasting from 1½ to 10 years.<sup>1</sup> The reported case presented as a smooth, firm, well defined swelling with an erythematous mucosa, slowly progressive over a period of 2 years.

Histologically, MEC contains three cellular elements: squamous cells, mucus-secreting cells and intermediate cells. The composition varies according to the grade of the tumour. According to the WHO classification (2005), they are classified into three grades: low, intermediate and high grade.<sup>2</sup> Low grade when cell atypia is minimal, cystic formation is prominent, and proportion of mucous cells is high; high grade when there is considerable pleomorphism and mucous activity and high proportion of quickly growing squamous cells, causing early pain, possible ulceration, bone resorption, lymphadenopathy, and even facial paralysis; and intermediate grade when the three types of cells are present but intermediate cells prevail.<sup>3</sup> The reported case showed predominant mucous cells with some intermediate type cells and was classified as low grade tumour.

Low grade tumours develop as a painless, slow-growing swelling, over several years, clinically mimicking a pleomorphic adenoma or other benign tumour, and rarely exceed 5 cm in size, whereas high-grade malignant tumours grow rapidly, are painful, associated with sensitivity or facial motor disorders, infiltrate adjacent tissues, and are associated with adenopathy and distant metastases, especially in the lung and bone.<sup>2</sup> The reported case was classified as low grade tumour.

The differential diagnosis for MEC is generally to distinguish it from other salivary gland tumours like adeno-squamous carcinoma, polymorphous low grade carcinoma, and squamous cell carcinoma. In MEC, the recurrence rate is approximately 25% and 10% in low grade lesions. Metastasis and survival rate are related to the histological grade and stage.<sup>1</sup>

The treatment of choice for minor salivary gland tumour is wide local resection with tumour-free margins. Resection margin status is one of the most important survival outcome prognostic factors that correlate strongly with both anatomical extent and histological type. The prognosis of MEC is mainly influenced by clinical stage, histological grade, and surgical margins, all strongly interrelated factors. In clinical practice only the low-grade subgroup without further negative prognostic features should be managed with surgery alone. In the intermediate-grade MEC group many patients will have additional negative prognostic factors (e.g. involved surgical margins or soft tissue invasion) that will be an indication for adjuvant radiotherapy.<sup>6</sup>

The reported case being classified as low grade, being well defined without any bony, adjacent structure and neck node involvement, surgery with wide local resection with 1.5 cm margin for marginal clearance was the treatment of choice and we found no recurrence on follow up.

**CONCLUSION**

The mucoepidermoid carcinoma of the minor salivary glands of the retromolar trigone presents a rare entity, with a clinical picture, prognosis, and therapeutic management, depending on the histological grade. The main therapy is surgical removal combined with radiotherapy essentially for the forms of high grade malignancy.

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