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CASE REPORT

Epithelial Myoepithelial Carcinoma of Palate: Consider, Contemplate and Confirm

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ABSTRACT

Diagnosing palatal swellings presents a challenge for clinicians due to the diverse etiologies, including developmental, inflammatory, reactive, and neoplastic origins. Among salivary gland tumors, epithelial-myoepithelial carcinoma (EMC) is an uncommon malignancy, constituting only 1–2% of salivary gland tumors, predominantly affecting the parotid gland. Its occurrence in minor salivary glands is exceptionally rare, accounting for less than 1% of cases. This report discusses a case of intraoral EMC arising from minor salivary glands of the palate, emphasizing the importance of accurate diagnosis and management to mitigate morbidity and mortality.

Keywords: Minor Salivary Gland Neoplasm; Epithelial Myoepithelial Carcinoma; Palatal Swellings

1 INTRODUCTION

First identified by Donath et al., epithelial-myoepithelial carcinoma was previously classified under various terminologies, including adenomyoepithelioma and clear cell adenoma.[(1)] It primarily affects the parotid gland, followed by the submandibular gland and minor salivary glands, with rare cases reported in sites such as the maxillary sinus, trachea, larynx, hypopharynx, and breast. The mean age of diagnosis is 60 years, with a slight female predominance, although cases have been documented in younger patients.[(2)] Clinically, EMC typically manifests as a painless, slow-growing mass; however, poorly differentiated forms may exhibit aggressive behavior with pain and nerve involvement.[(3)] Generally considered a low-grade malignancy, EMC has a favorable prognosis, with low recurrence rates and rare metastasis.[(4)] In the current article, a case of EMC of the palate is discussed with emphasis on differential diagnosis of palatal swellings in the light of literature.

2 CASE REPORT

A 77-year-old female, with no history of tobacco or alcohol use, presented with a swelling on the left side of the palate persisting for three weeks. The patient was unaware of its onset, and it gradually increased in size. The swelling was not associated with bleeding or pus discharge. Despite antibiotic and analgesic treatment, the lesion did not resolve. Clinical examination revealed a dome-shaped, well-demarcated swelling approximately 2 cm × 3 cm in size on the posterior hard palate, slightly toward the midline, with superficial ulceration [Figure 1]. The lesion extended from the rugae region to the junction of the soft and hard palate and laterally toward the gingival zone of the maxillary molars. The swelling was firm, non-pulsatile, and without endonasal expression or cervical lymphadenopathy. Based on the clinical presentation, a provisional diagnosis of minor salivary gland neoplasm was considered. The differential diagnoses considered in this case included palatal abscess, salivary gland neoplasms, reactive lesions such as necrotizing sialometaplasia, and

mesenchymal tumors.

Fig. 1: Dome shaped swelling on left side of the palate with superficial ulceration

CBCT imaging revealed a soft tissue mass with lobulated contours measuring 26.1 × 19.6 mm, exhibiting thinning of the hard palate and the nasal cavity floor [Figure 2]. Incisional biopsy demonstrated tumor cells arranged in nests, tubules, microcysts, and focal solid areas, suggestive of epithelial-myoepithelial carcinoma [Figure 3].

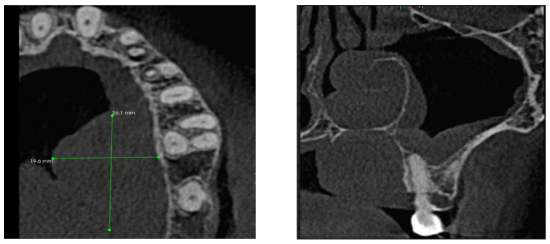

Fig. 2: Axial and coronal views of CBCT showing the extension of swelling

A wide local excision, including neck dissection, was performed. Histopathological analysis confirmed the diagnosis, with tumor cells exhibiting positivity for CK7, S100, and CD117, along with diffuse positivity for p63 and p40 in a subset of cells. Immunohistochemical and morphological findings corroborated the diagnosis of EMC.

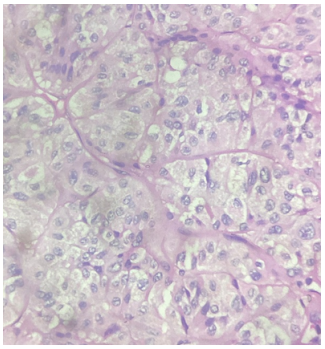

Fig. 3: Tumour cells of varying pattern including nests, tubules, microcysts and focal solid areas

3 DISCUSSION

Diagnosing palatal swellings requires careful clinical assessment due to the region's complex anatomical structure and the variety of potential pathologies.[5]

Palatal abscess was ruled out due to the absence of infection or inflammation. Pleomorphic adenoma, the most common minor salivary gland neoplasm of the hard palate, typically presents as a painless, slow-growing lesion, often without ulceration.[6] Malignant minor salivary gland tumors are more frequent in older individuals and can present with superficial ulceration, as seen in this case. Mucoepidermoid carcinoma, while common in intraoral minor salivary glands, often appears as a firm, fixed swelling, primarily affecting patients in their third to sixth decades of life.[7]

Necrotizing sialometaplasia is a benign, self-limiting condition affecting minor salivary glands, typically presenting as a nodular swelling that progresses to ulceration. Unlike EMC, it is more prevalent in middle-aged men.[8] Schwannomas, although rare in the oral cavity, typically present as slow-growing, painless nodules without ulceration unless traumatized.[9] Other differential considerations included vascular leiomyomas, rhabdomyosarcomas, and non-Hodgkin lymphomas, each of which has distinct clinical and histopathological features.[10]

The hard palate, with its rich presence of minor salivary glands, exhibits a unique distribution of neoplasms. Malignancies of the hard palate predominantly include minor salivary gland tumors (60.6%), followed by benign mesenchymal tumors (15.2%), squamous cell carcinoma (12.1%), malignant melanomas (6.1%), lymphomas (3.0%), and sarcomas (3.0%). EMC remains a rare entity, with fewer than 600 reported cases since its initial description in 1972.[11] Despite its classification as a low-grade malignancy, some morphologically low-grade EMCs demonstrate aggressive behavior.[12]

Histopathologically, EMC is characterized by well-defined tubules with a biphasic cellular arrangement: an outer layer of myoepithelial cells with clear cytoplasm encasing an inner layer of eosinophilic cuboidal epithelial cells. Immunohistochemistry plays a crucial role in confirming the diagnosis, with markers such as CK7, S100, p63, and CD117 aiding differentiation from other salivary gland tumors.[13]

While no standardized treatment protocols exist for EMC, surgical excision with clear margins remains the primary modality, often supplemented with adjuvant radiotherapy in selected cases to minimize recurrence risk.[14] The efficacy of chemotherapy remains uncertain. Studies have reported a recurrence rate of approximately 36.3%, with five- and ten-year survival rates of 93.5% and 81.8%, respectively.[15]

4 CONCLUSION

Minor salivary gland tumors, particularly EMC, remain rare but significant pathologies. Due to its architectural complexity and subtle clinical behavior, accurate diagnosis is essential. Further case studies are needed to enhance our understanding of EMC's demographic distribution, histopathological features, and optimal management strategies to improve clinical outcomes.

4.1 Conflict of Interest

No conflicts of interest to declare.

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