## **Case Report**

DOI: https://dx.doi.org/10.18203/2349-2902.isj20253051

# Clear cell myoepithelioma of the palate: a case report and comprehensive review

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Received: 08 July 2025 Accepted: 09 September 2025

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## **ABSTRACT**

Myoepitheliomas are a group of rare tumors of the salivary glands, primarily affecting the parotid gland. It accounts for less than 1% of all salivary gland tumors. The diagnosis of tumors of myoepithelial origin relies on evaluating the histologic features and is confirmed using immunohistochemical analysis. Vimentin and S-100 are non-specific but sensitive immunohistochemical markers of neoplastic myoepithelium. This is a unique case of a clear cell myoepithelioma of the hard palate. Diagnosis involved thorough histological examination and immunohistochemical confirmation, utilizing standard protocols. Our case showcases a rare occurrence of myoepithelioma, specifically the clear cell variant, in the hard palate. Histological analysis revealed large polyhedral cells with eosinophilic and clear cytoplasm and round to oval vesicular nucleus with small nucleoli. The cystic/ pseudo glandular spaces were filled with mucinous material. The results were further validated by immunohistochemical tests using Vimentin and S-100. This case highlights unusual presentation of myoepithelioma outside the parotid gland, emphasizing the importance of considering these tumors as differential diagnosis. Awareness of these atypical manifestations is crucial for accurate diagnosis and optimal patient management.

Keywords: Myoepithelioma, Hard palate, Salivary gland tumors, Clear cell, Rare

## INTRODUCTION

Palatal swelling may arise as a result of various etiological factors. The numerous minor salivary glands within the submucosal layer of the palate can contribute to the development of various pathologies. Salivary gland neoplasm accounts for approximately 3-5% of all the neoplasm of the maxillofacial region. Of these, 10-15% of all the minor salivary gland tumours are located in the palate. The minor salivary gland tumour either benign or malignant appears as a well-circumscribed lesion with a slow growth rate. Myoepitheliomas account for less than 1% of all salivary gland tumours. They are rare salivary gland neoplasms, usually slow-growing and asymptomatic. In 1943, Sheldon first coined the term

myoepithelioma to describe a group of tumours comprising of myoepithelial cells.

These cells are derived from ectodermal cells located between the basement membrane and the basal surface of acinar units.<sup>3</sup> Palate accounts for 21% of all myoepitheliomas.<sup>2</sup> Based on histological studies, myoepitheliomas display either non-myxoid (solid), myxoid (like pleomorphic adenoma), reticular or mixed growth patterns. The lesions are composed of spindle type (32.5%), hyaline type (7.5%), epithelial type (45%), clear type (2.5%) or mixed type (12.5%) tumour cells.<sup>4</sup>

Tumours with mild cytologic atypia were classified as myoepithelioma, whereas tumours with moderate to severe atypia were classified as myoepithelial carcinoma.<sup>5</sup>

The malignant lesions should be differentiated from their benign counterparts by the presence of infiltrating growth, areas of necrosis, high mitotic rate, cytologic atypia and cellular pleomorphism.

## **CASE REPORT**

A 47-year-old patient presented with a complaint of growth in her right side of the palate for 6 months. The lesion was initially small in size and gradually progressed to the presenting size. The lesion was not associated with any bleeding/discharge and was non-tender. There were no co-morbidities reported. There was no facial asymmetry (Figure 1).



Figure 1: Extra-oral appearance with no facial asymmetry.

Examination revealed a solitary, partially pedunculated growth was seen arising from the right side of the palate and crossing the midline (Figure 2). The lesion had a smooth surface, was not covered with slough, was soft to firm in consistency, nonulcerated and was noncompressible and non-fluctuant. Regional lymphadenopathy was absent.



Figure 2: Intra-oral palatal solitary lesion.

Computed tomography (CT) revealed a lobulated, well-circumscribed mass arising from the right side of the palate measuring  $3.6 \times 2.4 \times 1.2$  cm (Figure 3). The mass was seen extending from the right maxillary alveolus up to the soft palate posteriorly, closely abutting the root of the

pterygoid plates (Figures 4 and 5). There was mild bony remodelling noted without any areas of bony erosion.



Figure 3: Coronal sections of CT showing adherence to palatal bone.

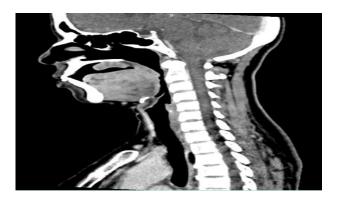


Figure 4: Sagittal section showing the posterior extent of growth.



Figure 5: Axial section showing the extension of the lesion.

An incisional biopsy of the lesion was performed. Histological sections showed sheets of tumor cells composed of large polyhedral cells having eosinophilic and clear cytoplasm and round to oval vesicular nucleus with few small nucleoli (Figure 6).

The few tumor cells were plasmacytoid with the eccentrically placed nucleus. Cystic/pseudo glandular spaces filled with mucinous material were frequent. In

between sheets of cells, hyalinized fibrous septae were seen (Figure 7). Immunohistochemical staining showed tumor cells positive for P63, Ki67 (8%), CK7 and Vimentin and occasional cells showed positivity for S-100. Due to the borderline percentage of Ki67, wide resection of the lesion was planned which was carried out under general anesthesia. The underlying bone was excised, and Oro-antral communication was noted.

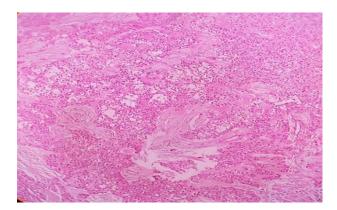


Figure 6: Photomicrograph showing sheets of tumor cells composed of large polyhedral cells having eosinophilic and clear cytoplasm and round to oval vesicular nucleus (H & E stain, 10X magnification).

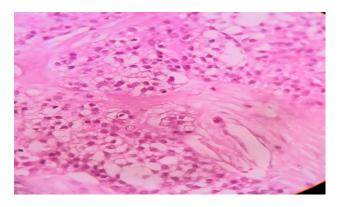


Figure 7: Photomicrograph showing few plasmacytoid tumor cells with eccentrically placed nucleus and hyalinized fibrous septa in between sheets of cells (H & E stain, 40X magnification).



Figure 8: Follow up of 6 months with reduced size of Oro antral fistula.

The closure of the communication was planned at a later stage to monitor for local recurrence. The patient reported for follow-up for 6 months with a feeding plate obturator without any signs of recurrence (Figure 8). The patient was lost for follow-up later.

## **DISCUSSION**

Myoepitheliomas are benign, rare salivary gland neoplasms. Batsakis et al stated, "tumors that are completely devoid of epithelial cells, merit the designation myoepithelioma". Luna et al defined them as "well-circumscribed and encapsulated lesions with a hyalinized stroma which lacks the myxoid or chondroid appearance of the mixed salivary gland tumors". Sciubba and Brannon stated that these tumors are "composed entirely of myoepithelial cells". Most of these tumors are in the parotid gland, while others may occur in the submandibular or accessory glands of the oral cavity. The clinical behaviour of the myoepitheliomas is like other benign salivary gland tumors, slow-growing and usually asymptomatic. It is most seen in the 3rd-4th decade without gender predilection.

Myoepithelial cells are routinely found in tissues with a secretory function such as salivary glands, lacrimal and sweat glands, prostate and breasts and are believed to have contractile properties. These cells are flattened and spread over the acini as long cytoplasmic processes.<sup>4</sup> They are responsible for the ejection and propagation of secretions from the acini through the ductal network.<sup>9</sup> These ectodermal derived cells contain cytokeratin, polypeptides and actomyosin filaments. Initially, it was considered that these cells are localized to acini and intercalated ducts of the salivary glands. However, recent immunohistochemical evidence shows that these cells may also be present on outer aspects of intra- and interlobular striated ducts.

Due to the various phenotypic and ultrastructural modifications by the myoepithelial cells of salivary gland tumors, the morphologic spectrum of myoepitheliomas has been expanded. The spindle cell type contains multiple nodules separated by a narrow fibrous band. These nodules are comprised of closely packed, uniform spindle cells. The spindle cell in myoepithelioma with reticular patterns have cells that are plump and more irregular while those in the myxoid variant are smaller with limited cytoplasm and "streaming alignment". 4 The plasmacytoid or hyaline type consists of cells that are oval to polygonal in shape with eccentric nuclei of epithelial cells arranged in nests or cords. These tumor cells have limited endoplasmic reticulum, lack Golgi complex and the hyaline cells exhibit a halo of tonofilaments around the nucleus. Plasmacytoid myoepithelioma is differentiated from plasmacytoma by larger cell size and absence of both cytoplasmic immunoglobins and perinuclear halos. 9 In the epithelial type, the cells are polygonal with an abundant amount of cytoplasm. In the myxoid variant of the epithelial and plasmacytoid type, the cells are arranged in small groups separated by varying amounts of proteoglycans. Clear cell type is the only solid myoepithelioma comprising tumor cells with faint eosinophilic or non-staining cytoplasm. Accumulation of glycogen and widened intercellular spaces results in a clear cytoplasmic appearance. In the mixed cell type, two different patterns of cellular differentiation where neither is predominant or the second component accounts for more than 10% of tumor cells. Out of these 5 main histologic subtypes of myoepitheliomas, the spindle cell variant is the most common.<sup>4</sup> Douglas reviewed "signet ring cell" adenocarcinomas of the salivary gland that would stain for myoepithelial markers and contained intracellular mucin materials and classified them as mucinous epitheliomas.<sup>10</sup>

Pleomorphic adenoma is the main differential diagnosis of myoepitheliomas. They are considered to be two different forms of the same entities. Pleomorphic adenoma affects women between 30-60 years and presents as a slow-growing painless mass. Myoepitheliomas are almost entirely composed of myoepithelial cells, whereas variable amounts are seen in pleomorphic adenoma. Unlike pleomorphic adenoma, myoepitheliomas lack ductal epithelial differentiation. If the tumor comprises less than 5% of ductal and acinar components it is considered as myoepithelioma. The presence of ducts in myoepithelioma could be due to two reasons, passive incorporation of normal salivary structures into the tumor during growth and stem cells have a high incidence of innate ability to form both myoepithelial and duct cells.

Verocay bodies have been observed in some myoepithelioma and are not unique nerve sheath tumors. Nerve sheath tumors can be differentiated from myoepithelioma by the presence of Schwann cells surrounded by a basement membrane. The incidence of leiomyoma occurring in the palate accounts for 18.9% of cases. They are usually asymptomatic with smooth surface and is typically submucosal, the age of onset ranges from 40-49 years. Leiomyomas can be differentiated by myoepitheliomas, the former shows myofilaments, dense bodies and pinocytotic vesicles. Pyogenic granulomas are reactive lesions, that appear as a smooth or lobulated exophytic swelling with a pedunculated or sessile base. Follicular hyperplasia is often seen in older females and appears as a firm, nonulcerated, painless mass without bone involvement.<sup>12</sup> The confirmed diagnosis for any soft tissue lesion can be concluded only by histopathological evaluation.

Various immunohistochemistry markers have been for myoepitheliomas. S-100 proposed protein immunoreactivity has been used to assess myoepithelial differentiation. They are usually present in neoplastic myoepithelial cells but not normal salivary gland myoepithelial cells. Vimentin is one of the indicators of neoplastic myoepithelial cell differentiation. Ki-67 labelling index helps assess cellular proliferation and to distinguish between benign and malignant myoepitheliomas. Its expression is higher in malignant

tissues with poorly differentiated tumor cells, compared to normal cells. Ck-7 is positive for all malignant salivary gland tumors and differentiates squamous cell carcinoma from primary salivary gland tumors.

The imaging findings of myoepithelioma are highly non-specific. Ding et al investigated myoepitheliomas of the parotid gland and observed that they appeared as unilocular, rounded tumors with smooth contours, displaying capsule on T2W and contrast enhanced T1W imaging, exhibiting homogenous signal intensity on CT and MRI.<sup>13</sup>

Myoepitheliomas may occasionally infiltrate locally and metastasise. Even though the majority of myoepithelioma is benign or low-grade neoplasm, there is an approximate 20% risk for local recurrence.<sup>5</sup> The recommended management of myoepithelioma is wide excision of the lesion. A margin of 0.5 cm is recommended to reduce the risk of recurrence.

### **CONCLUSION**

In conclusion, this case report presents a rare instance of a clear cell myoepithelioma of the palate. The patient was successfully treated with complete surgical excision, confirming that histopathological examination with immunohistochemistry is the definitive method for diagnosis. Given the benign nature and complete resection, the patient's post-operative course was uneventful with an excellent prognosis. This case highlights that while clear cell tumors can be diagnostically challenging, awareness of myoepithelioma as a differential diagnosis is crucial for appropriate surgical management and follow-up.

Funding: No funding sources Conflict of interest: None declared Ethical approval: Not required

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Cite this article as: Anehosur V, Kotian DB, Anand J, Kumar K, Kumar N. Clear cell myoepithelioma of the palate: a case report and comprehensive review. Int Surg J 2025;12:1870-4.