

## A Rare Case Report of Plasmacytoid Myoepithelioma of Hard Palate

Swarna YM<sup>1\*</sup>, Ali IM<sup>2</sup> and Rajeshwari G Annigeri<sup>2</sup>

<sup>1</sup>Faculty of Dentistry, MAHSA University, Damansara Heights, Kuala Lumpur-50459, Malaysia

<sup>2</sup>Department of Oral Medicine and Radiology, College of Dental Sciences, Davangere-577004, Karnataka, India

### Abstract

**Aim:** Myoepithelioma of salivary glands are extremely rare, comprising approximately only 1-1.5% of all salivary gland tumors.

**Background:** Minor Salivary gland tumors are infrequent, representing 10-15% of all salivary neoplasms. Their frequent location is parotid gland & myoepithelioma from palatal salivary glands is considered as a rare entity. Yet again, the Plasmacytoid Myoepithelioma from palatal salivary glands is sporadic, till date it has been reported merely 15 cases.

**Case description:** Here we report a 51 years old female patient with a slow growing palatal swelling. Cytological features of FNAC were suggestive of Myoepithelioma. The patient underwent wide surgical excision of lesion and after a month patient was followed up wherein oronasal communication was existing, subsequently patient was provided with the maxillary obturator.

**Conclusion:** Clinically, its presentation may resemble pleomorphic adenoma, which hampers the correct diagnosis.

**Clinical relevance:** This case report reinforces the need to alert dentists to include Myoepithelioma in the differential diagnosis of palatal swellings in elderly patients.

**Keywords:** Plasmacytoid; Myoepithelioma; Palatal swellings

### Introduction

Salivary gland neoplasms with exclusively of myoepithelial cells are unusual and intriguing [1,2]. Myoepitheliomas are rare tumor of salivary gland constituting less than 1% of all salivary gland tumors [3]. Sheldon, first used the term "Myoepithelioma" in 1943 and was included in the 1991 revised WHO classification of salivary gland tumors [3,4].

The growth patterns may be solid, myxoid or reticular and the component cells may be spindle shaped, plasmacytoid, hyaline, clear or epitheloid [5]. The plasmacytoid myoepithelioma from palatal salivary glands is considered as a very rare entity [6]. Palatal masses often lead to a diagnostic dilemma because of their similar presentations.

Here, we report a case of Plasmacytoid Myoepithelioma of palate which resembles pleomorphic adenoma of palate.

### Case Report

A 51 year old lady reported to our department with the slowly growing painless palatal mass of 2 years duration which was insidious in onset. Patient gave a history of discomfort while chewing and swallowing since 6 months and negative history of fever, trauma and any other swelling in the body.

Medical records suggested patient was known hypertensive since 2 years, but she was not on medication. During examination, patient was found to be in good general and physical health.

On extra-oral examination, no abnormality detected except palpable, firm and non-tender right and left solitary submandibular lymph nodes, whereas intraoral examination (Figure 1) revealed a well-defined 3×4 cm sized ovoid solitary swelling in the palate extending anteroposteriorly mesial of 23 to distal of 28 and mediolaterally from palatal gingival margin of 23-28 to 2 cm away from the palatal gingival margin of contralateral side, with red color change indicating engorged vessels and without any change on the surface of swelling. To palpate, swelling was firm and non-tender. The adjacent teeth 27, 28 were

mobile and were assigned grade II to grade III mobility respectively.

Considering history and clinical examination, case was provisionally diagnosed as "Pleomorphic Adenoma of hard palate".

Occlusal and Para nasal radiographs (Figures 2 and 3) revealed no abnormality. FNAC was done as a part of investigation and cytological features were suggestive of Myoepithelioma. Results of hematologic, biochemical investigations were within normal limits and HbsAg, HIV-1 & 2 tests were also negative (Table 1).

The patient underwent wide surgical excision along with a rim of surrounding normal tissue under general anesthesia in a private



Figure 1: Pre-Operative Intra Oral Photograph.

\*Corresponding author: Swarna YM, Faculty of Dentistry, MAHSA University, Damansara Heights, Kuala Lumpur – 50459, Malaysia, Tel: +60183279785; E-mail: drswaru@gmail.com

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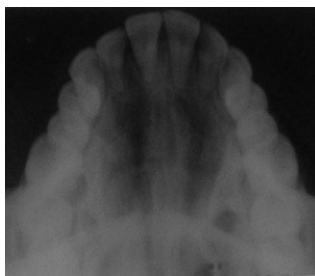


Figure 2: Pre-Operative True Occlusal Radiograph.



Figure 3: Pre-Operative Para nasal Sinus view.

Case	Author	Age (years)	Sex	Location	Course (months)	Outcome
1.	Politi et al. [2]	44-year old	Female	Right parotid gland	6 months	No recurrence
2.	Rastogi et al. [3]	33-year old	Male	Palate	8 months	No recurrence
3.	Ferri et al. [5]	81-year-old	Female	Right cheek	2-years	No recurrence
4.	Cuadra et al. [6]	28 year old	Female	Swelling of hard palate	One year and a half	No recurrence
5.	Sperandio et al. [9]	42 year old	Female	Left side of soft palate	6 months	No recurrence
6.	Zormpa et al. [10]	29-year-old	Female	Hard and soft palate	2-years	No recurrence
7.	Reddy et al. [11]	12-year old	Female	Palate	6 months	No recurrence
8.	Agarwal et al. [12]	40-year-old	Female	Palate	6 months	No recurrence
9.	Yadav et al. [13]	40-year-old	Male	Palate	4 years	No recurrence
10.	Medici et al. [14]	70-year old	Female	Palate	6 months	No recurrence
11.	Nara et al. [15]	60 years old	Male	Soft palate	2 years	No recurrence
12.	Santos et al. [16]	15 years old	Male	Hard palate	1 year	No recurrence
13.	Açikalin mf et al. [17]	32-year-old	Male	Soft palate	Unaware of the swelling	No recurrence
14.	Sethi et al. [18]	45 year old	Female	Junction of hard and soft palate	One and half year	No recurrence
15.	Sayed et al. [19]	57 year old	Female	Hard palate (sinonasal cavity)	3 years	No recurrence

Table 1: Showing distribution of 15 cases of Myoepithelioma, according to patient age, sex, location, duration of the lesion and treatment outcome.

clinic. Patient was followed up after 3 weeks, wherein inflammation and persistence of oronasal communication was seen (Figure 4). Histopathological examination, revealed predominantly proliferation of plasmacytoid cells arranged in discrete & clusters with eccentrically situated nuclei having abundant cytoplasm. Few clusters consist of round to polygonal cells & few were spindle to epitheloid type with elongated nuclei & eosinophilic cytoplasm (Figure 5). Features were suggestive of Plasmacytoid Myoepithelioma of minor salivary gland in the hard palate.

After a month patient was followed up wherein oronasal communication was existing, subsequently patient was provided with the maxillary obturator (Figure 6).

## Discussion

Minor salivary gland tumors are infrequent, accounting for 10-15% of all salivary gland neoplasms and are primarily located in the palate (50%), lips (15%), cheek mucosa (12%), tongue (5%) and floor of the mouth (5%) [1].

Myoepitheliomas are rare tumor of salivary gland constituting less than 1% of all salivary gland tumors [3]. Myoepithelial cells are thin



Figure 4: Post -Operative Intra Oral Photograph after 3 weeks.

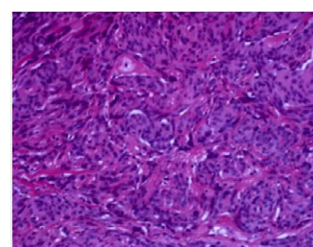


Figure 5: Histopathology.



Figure 6: Post-Operative Intra Oral Photograph with Obturator.

and spindle shaped and situated between the basement membrane and epithelial cells and these are the normal constituents of major and minor salivary glands, having the contractile properties, assisting in the secretion of saliva [3].

Salivary gland neoplasms that frequently contain myoepithelial cells are Pleomorphic Adenoma, Adenoid-cystic carcinoma, Epithelial-Myoepithelial carcinoma of intercalated duct origin [5].

Myoepitheliomas appear to be rare and this contrast with the active role of myoepithelial cells in the histogenesis of several types of salivary gland tumors [2].

The most common location for myoepithelioma of the head and neck region are the parotid gland (40%) and the palate (21%) [2]. This neoplasm seems to result from a monoclonal proliferation of myoepithelial cells, which may partially differentiate into spindle-shaped or plasmacytoid cells. Their histogenesis from myoepithelial cells is reflected in their most frequent location that is the parotid gland, wherein myoepithelial cells are more common [7].

The mean age at presentation of benign Myoepithelioma is about 50 years and the sex incidence is roughly equal. Most patients complain of a well-circumscribed mass, usually 10-50 mm in diameter in either major or minor salivary glands [4].

Myoepithelioma generally shows several cellular patterns namely spindle, plasmacytoid, epitheloid, clear cell. Growth patterns namely solid, myxoid, reticular may be seen. Myoepitheliomas of the minor salivary glands tend to be composed of plasmacytoid cells and those of the parotid; it may be epitheloid or spindle cells. The clear cell variant can occur in both major and minor salivary glands, but is relatively rare [4].

The rarity of Myoepithelioma makes it a surprise diagnosis; identifiable only histopathologically. Its clinical differential diagnosis usually consists of the more common salivary gland tumors. In the parotid region, Pleomorphic Adenoma, Warthin tumor, Basal cell adenoma, Adenoid cystic carcinoma or Mucoepidermoid carcinoma would be appropriate. In oral sites, the same differential would generally prevail except Warthin tumor and the canalicular adenoma which would be substituted for the basal cell adenoma [7].

Myoepitheliomas frequently mistaken for cellular Pleomorphic Adenoma, because both the tumors consist of abundant myoepithelial cells [8]. It has been proposed that if the neoplasm contains <5% ductal and acinar components, it must be named myoepithelioma and if there is ductal predominance, pleomorphic adenoma diagnosis should be established [3].

Immunohistochemically, a large number of markers have been used for establishing the diagnosis of myoepitheliomas, which includes Muscle Specific Actin (MSA), a marker of myogenous differentiation and epithelial filaments of cytokeratin [3].

With Immunohistochemical techniques, myoepithelial cells stain positive for cytokeratin, MSA, occasionally express S-100 protein and Glial Fibrillary Acidic Protein (GFAP). The neoplastic myoepithelial cells consistently demonstrate cytokeratin S-100 and MSA immunoreactivity whereas reactivity for vimentin and GFAP is more variable [2].

Biologically, in most cases myoepitheliomas are benign, but occasionally infiltrate locally and metastasize. The prognosis of myoepithelioma would appear to be good, provided surgical excision is complete [5].

In conclusion, Myoepithelioma of salivary glands are unusual tumors with plasmacytoid variants still sporadic. Because of their infrequency variable growth patterns and often because of increased cellularity they can be misdiagnosed. However, ultra structural study of myoepithelial cells establishes fine structural criteria on which positive identification could be made. Therapy should be directed towards complete surgical extirpation and close follow up is mandatory for all cases.

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