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CASE REPORT

Myoepithelioma Of Palate- A Case Report And Review Of Literature

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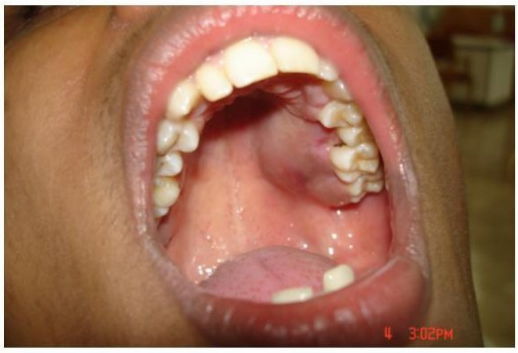
Introduction

Myoepitheliomas are rare tumor of salivary glands constituting less than 1% of all Salivary gland tumors [22]. Myoepithelial cells are situated between basal lamina and acinar ductal cells of salivary glands and other exocrine organs [20]. Sheldon [21] first used the term "Myoepithlioma" . It occurs mainly in parotid gland and is composed of myoepithelial cells [19], [22], [25].Salivary gland neoplasms that frequently contain myoepithelial cells are Pleomorphic Adenoma, Adenoid Cystic Carcinoma, and Epithelial-Myoepithelial Carcinoma of the intercalated duct origin[6]. These cells are difficult to identify in routine histological examination or electron microscopy and immunohistochemical examination is essential to establish the diagnosis of this rare entity [1], [8]. We report here a case of plasmacytoid myoepithelioma of hard palate with histological confirmation together with a review of literature.

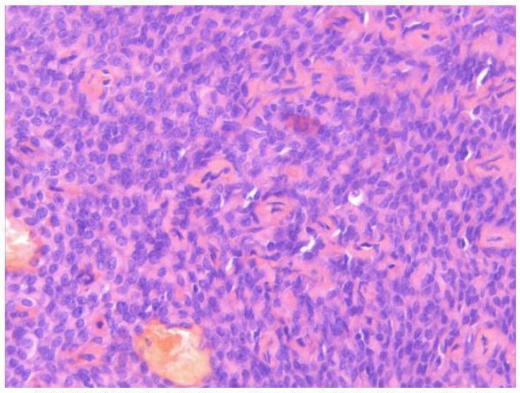
Case Report

A 33-year old man presented with a palatal mass of 8 months duration, insidious in onset, progressive in nature and not associated with any pain or pus discharge. Deglutition was normal. The overlying mucosa was normal in color and texture, firm, well-circumscribed and about 4x2cm in diameter [Table/Fig 1]. No regional lymphadenopathy and systemic problems were reported. Results of physical examination were normal, with no signs of metastases. Radiograph depicted no bony abnormalities. A clinical diagnosis of minor salivary gland tumor was made. Wide surgical excision of the lesion was performed. A 1-year follow-up showed no evidence of recurrence.

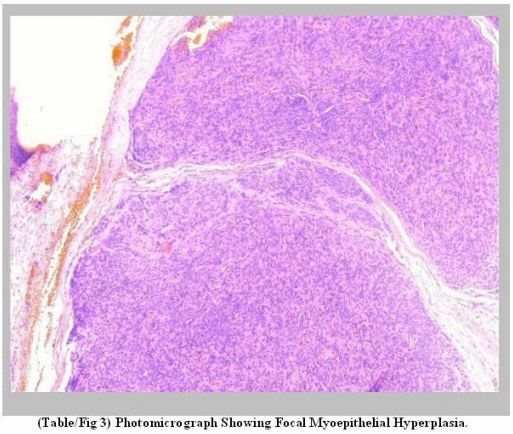
Histologically, the tumor is composed of closely packed nodules containing sheets and islands of neoplastic plasmacytoid myoepithelial cells with round to oval eccentric nuclei with abundant eosinophilic cytoplasm and focal cribriform pattern, surrounded by dense eosinophilic collagenous stroma containing residual ducts and acini of minor salivary glands with focal myoepithelial hyperplasia [Table/Fig 2], [Table/Fig 3]. Immunohistochemistry was not done due to lack of facilities; the diagnosis was based on histological findings.



(Table/Fig 1) Intraoral View Showing A Well-Circumscribed Swelling In The Posterior Part Of The Hard Palate On The Left Side.



(Table/Fig 2) Histologically Tumor Shows Closely Packed Nodules Containing Sheets And Islands Of Neoplastic Plasmacytoid Myoepithelial Cells With Round To Oval, Eccentric Nuclei.



(Table/Fig 3) Photomicrograph Showing Focal Myoepithelial Hyperplasia.

Discussion

Myoepithelial cells are a normal constituent of major and minor salivary glands and are believed to have contractile properties, which assist in the secretion of saliva. They are also found in breast, tracheo-bronchial and sweat glands. They are plentiful in salivary acini and intercalated ducts, but much less so in larger excretory ducts of major glands. Microscopic examination shows that myoepithelial cells are thin and spindle shaped and situated between the basement membrane and epithelial cells. Myoepithelioma is a benign neoplasm of salivary glands that represents 1.5% of all salivary glands neoplasms [1]. Their most frequent location is parotid glands. No gender predilection has yet been reported, and highest frequency is observed in the third decade of life [1].

On rare occasions, myoepitheliomas arise in intraoral minor salivary glands [25] and a number of single cases with sufficient documentation have been published. In a review of literature 13 cases of ultrastructurally or immunohistochemically confirmed myoepithelioma of minor salivary gland origin were found [Table/Fig 4]. The average age of patients with intra-oral myoepithelioma was reported as 30-40 years. Most frequent site of origin was palate (10 of 13 cases). The tumor generally presents a benign clinical course; common presentation being a non-painful swelling or mass. The masses ranged from 1.5-4cm in their greatest dimension. In this reported case, size of the swelling was 4x2cm and gross findings were described as a well-circumscribed and solid encapsulated mass without ulceration. Twelve of the earlier reported cases were of plasmacytoid type.

(Table/Fig 4) Reported Cases Of Myoepithelioma Of Minor Salivary Gland Origin.

Author	Age/Sex	Site	Cell type
Scubba and Brannon[22]	##	##	Plasmacytoid
Kahn and Schoub[15]	17/F	Hard Palate	Plasmacytoid
Luna et al.[16]	30/F	Hard Palate	Spindle
Stromeyer et al.[24]	14/M	Gingiva and Anterior Maxilla	Cytologic Pleomorphism
Scubba and Goldstein[23]	22/M	Palate	Plasmacytoid
Crissman et al.[7]	81/M	Parotid	Plasmacytoid
Nesland et al[18]	18/F	Soft Palate	Plasmacytoid
Barnes et al[3]	24/F	Hard Palate	Plasmacytoid
Thompson et al.[26]	23/M	Oral Floor	Plasmacytoid
Enomoto et al.[11]	57/F	Soft Palate	Plasmacytoid
Ellyn and Gnepp[10]	8/F	Soft Palate	Plasmacytoid
Kawabe et al[14]	53/M	Soft Palate	Plasmacytoid
Harusachi et al.[13]	42/F	Hard Palate	Plasmacytoid
Present Case	33/M	Hard Palate	Plasmacytoid

Clinical and biologic behavior of this tumor is unknown. Malignant counterpart of

myoepithelioma has also been described which is a locally aggressive tumor with metastatic potential (especially to lungs, inguinal lymph nodes, liver, lumbar spine, pleura, peritoneum and skin) [1],[7],[21],[24] and with a strong tendency to recur locally. Signs of malignancy include ulceration, pain, rapid and sudden growth and bone infiltration. Treatment includes surgery, radiation therapy or a combination of both. Radiation therapy is supposed to be used only in cases where surgery is not feasible. Since, the biologic behavior of tumor is unknown, close follow-up is essential. In this reported case, there were no signs of malignancy and tumor was treated by wide excision without any recurrence until after 1 year of follow-up.

Myoepithelioma generally shows several cellular patterns namely; spindle, plasmacytoid, epithelioid and clear cell [9],[25].The plasmacytoid cells have a plasma cell-like appearance but are much larger and do not show the typical "nuclear halo". They are called as "hyaline cells"[15]. These plasmacytoid-type tumor cells tend to occur more frequently in the oral cavity, especially palate, whereas spindle cell types have been reported more in parotid gland [9], [22].

Although specific diagnostic criteria to differentiate myoepithelioma from pleomorphic adenoma have been proposed, controversies still exist. Sciubba and Brannon [22] consider myoepithelioma as a subtype or final spectrum of pleomorphic adenoma, because of its biological behavior and distribution. On the other hand World Health Organization classifies myoepithelioma as an independent entity [1]. It has been proposed that if the neoplasm contains less of 5% of ductal and acinar components, it must be named myoepithelioma [1], [7].Seifert distinctly separated myoepithelioma by showing no ductal differentiation and absence of chondroid or myxochondroid foci. To establish specific percentages like 5% or 10% of determinate, histological components could be subjective for each observer. However, this quantitative parameter constitutes the diagnostic criteria accepted to establish a differential diagnosis between pleomorphic adenoma rich in myoepithelial cells and plasmacytoid myoepithelioma. On the other hand, a diagnostic differentiation based on cellular predominance could be a better alternative for consideration. We suggest that, myoepithelial predominance should be a diagnostic feature for myoepithelioma and if there is ductal predominance, a pleomorphic adenoma diagnosis should be established. However, until this suggestion receives confirmation or rejection by other research groups, the percentage of ductal structures will continue to be considered as the principal diagnostic parameter for differentiation of plasmacytoid myoepithelioma of salivary glands from pleomorphic adenomas rich in myoepithelial cells.

Electron microscopic and immunohistochemical examinations are useful for accurate identification and characterization of myoepithelial cells. Ultrastructural findings indicate that myoepithelial cells often contain desmosomes, myofilaments in the presence or absence of dense bodies, tonofilaments, and basal lamina [3],[10],[11],[15],[16],[18],[23],[26],[28] Immunohistochemically, various markers have been proposed for diagnosing myoepithelioma which includes, MSA, a marker of myogenous differentiation and epithelial filaments of cytokeratin[5]. But there is considerable variation in the tumor cell expression of MSA; spindle cells react strongly to MSA whereas plasmacytoid cells generally show negative [8], [9], [27], [28],[29],[30] response which is attributed to neoplastic modified myoepithelial cells that are not fully differentiated [1],[8],[12].

S-100 protein immunoreactivity has also been used considerably in the past as an evidence of myoepithelial differentiation, but Takai [27] have proved that this is not

the case since it is present in neoplastic salivary gland cells.

Vimentin, an intermediate filament protein of fibroblasts and other mesenchymal cells, is one of the earliest indicators of neoplastic myoepithelial cell differentiation [8], [17] Recently, Glial fibrillary acidic protein, is also found to be potentially expressive in myoepithelioma.
Both Vimentin and Glial fibrillary acidic protein are more commonly used As markers for myoepithelioma than MSA. [1], [8], [12].

Conclusion

Plasmacytoid myoepitheliomas are rare salivary gland neoplasms. Cytologic pleomorphism is a frequent histological feature, which has led to questionable diagnosis of malignancy in some cases. Caution is advocated in the evaluation of a salivary gland neoplasm with such features, and confusion with primary squamous cell or undifferentiated carcinoma of the salivary glands should be avoided with use of appropriate immunohistochemical and electron microscopic examinations. Patients with cytologically pleomorphic lesions should be assessed for evidence of metastases. Therapy should be directed towards complete surgical extirpation and close follow-up is mandatory for all cases.

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