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Case report

PLASMACYTOID MYOEPIITHELIOMA OF MINOR SALIVARY GLANDS: A RARE CASE REPORT

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ABSTRACT

Myoepithelioma of the salivary glands is a rare benign neoplasm with incidence of less than 1% of all salivary gland neoplasms. The most common site is the parotid gland followed by minor salivary glands. These tumors occur at any age with peak incidence in the third & fourth decade. Here we report a case of plasmacytoid myoepithelioma of the minor salivary glands of soft palate which was conclusively diagnosed on FNAC and further confirmed by histopathological studies. The rarity of the tumor and the site has been emphasized.

Key words: Myoepithelioma, Minor salivary gland, Plasmacytoid variant, Soft palate.

INTRODUCTION

Myoepithelioma is believed to be rare entity in the tumours of salivary glands with less than 100 cases reported in the literature. It was first described by Sheldon in 1943 and was considered as variant of pleomorphic adenoma.^[1] But now-a-days most authors consider myoepithelioma as a distinct pathological entity, composed entirely of myoepithelial cells behaving much more aggressively than pleomorphic adenoma. Myoepithelioma arises from myoepithelial cells which are usually present in ductal epithelium of secretory glands like salivary, sweat and mammary glands.^[2] Myoepithelial cells are characterised by intracytoplasmic myofilaments, intercellular desmosomes and myogenic markers.^[3] Histopathologically there are five variants; spindle cell, plasmacytoid, epithelioid, clear cell and mixed variant. Spindle cell variant is the most common followed by plasmacytoid variant. Majority of myoepitheliomas present as painless, slow growing, well circumscribed, smooth surfaced

tumors. They are well capsulated and rarely metastasize. However recurrences have been described.^[3]

CASE REPORT

A 60 year old male patient came to the OPD of ENT department with swelling in the oral cavity since 2 years. On examination swelling was noticed on the left side of soft palate, measuring about 4 x 3.5cm, well circumscribed, smooth surfaced. Overlying mucosa was intact and not traumatized (Fig-1). There was no evidence of cranial nerve and lymph nodal involvement. The past history and family history were not relevant. Routine blood and biochemical investigations of patient were within normal limits. Patient was advised FNAC which showed high cellularity consisting of plasmacytoid cells in sheets, clusters and singles on a background of myxoid stromal fragments (Fig-2). Surgical excision was done (Fig-3) and surgical specimen was sent for

histopathological examination. Histopathological examination revealed a solid tumor consisting of plasmacytoid cells in nests, islands and cords separated by scanty myxoid stroma. There were no ductal or glandular elements, as well as no atypia or necrosis in the sections studied. Thus, confirming the diagnosis of plasmacytoid myoepithelioma. (Fig-4)

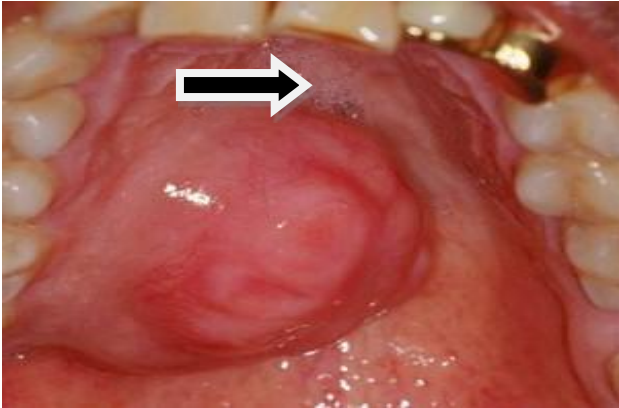


Fig 1: Tumour measuring 4x3.5cm on the soft palate.

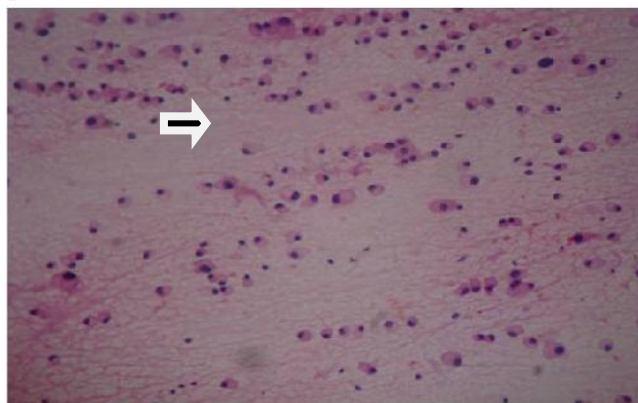


Fig 2: FNAC Tumour cells arranged in sheets. Plasmacytoid myoepithelial cells with rounded nuclei eccentrically placed with eosinophilic hyaline cytoplasm. Background showing myxoid stromal elements (H&E; 100x).



Fig 3: Intra-operative tumour mass removed along with capsule from soft palate

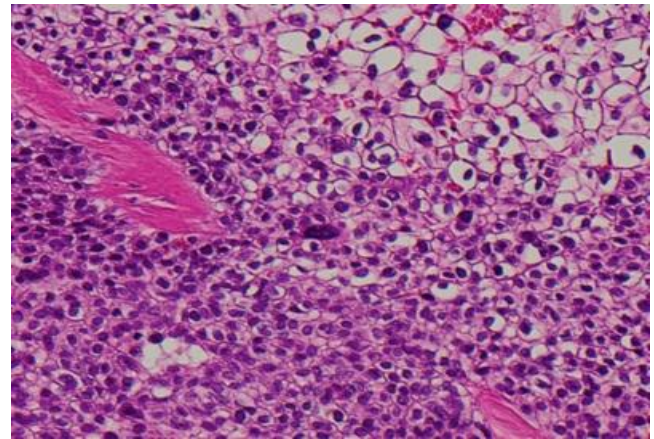


Fig 4: Histopathological study (H&E,100x) showing tumour tissue arranged in diffuse sheets. Most of the tumour cells have eccentrically placed nuclei with hyaline eosinophilic cytoplasm (i.e. plasmacytoid cells). Also seen spindle shaped cells and few foci of clear cell change. Stroma shows hyalinised collagen fibres.

DISCUSSION

Myoepithelioma is rare benign neoplasm of salivary glands. Among its four sub-types spindle cell type is most common (seen in 70% cases) where as plasmacytoid cell type is seen in only 20% cases. Plasmacytoid cell type is more common in major salivary glands.^[4] Therefore plasmacytoid myoepithelioma in minor salivary glands is a rare entity. The biggest series published on myoepithelioma is by Scuibba and Brannon who presented 23 cases of myoepithelioma of salivary glands (both major and minor).^[3] According to literature review only 14 cases of plasmacytoid variant of myoepithelioma affecting minor salivary glands of palate have been reported.^[5] Age distribution ranged from 3rd decade to 9th decade, with mean age of 53 years. No sex predilection has been described.^[2] Myoepithelioma must be differentiated from pleomorphic adenoma by absence of chondroid or osteoid changes in the matrix and absence of inconspicuous ductal differentiation.^[1] Benign myoepithelioma can be differentiated from malignant myoepithelioma by absence of solid pattern, infiltrating growth, necrotic areas, mitotic figures, hyperkeratotic nuclei, cellular polymorphism, cellular atypia and metastases.^[6] Malignant myoepithelioma has also been identified by cell proliferation index (>10% highly suggestive of

malignant behavior). Basal membrane globule surrounded by hyperkeratotic myoepithelial cells goes in favor of malignant myoepithelioma.^[1] Differential diagnoses of plasmacytoid myoepithelioma include myoepithelial cell predominant pleomorphic adenoma, plasmacytoma, lymphoma, skeletal muscle and rhabdoid tumors. Presence of myxohyaline stroma and cohesive clusters of plasmacytoid cells favor myoepithelioma over other diagnosis. Absence or less than 5% of epithelial cells showing ductal or acinar formation and absence of chondroid stroma helps in differentiating it from pleomorphic adenoma. In our case myxohyaline stromal fragments were noted and inconspicuous ductal or acinar pattern helped us to distinguish it from pleomorphic adenoma.^[7] In immuno histochemical study myogenic markers like CK-14, CK-18 & 19 is expected to be positive in plasmacytoid variant. Surgical excision with margin (few mm) of normal tissue is the treatment of choice. Recurrences are rare. According to Sciubba & Brannon, who followed-up 16 cases out of 23 over a period of 1 year, found recurrence only in one case.^[3] Recurrences can be picked up by regular follow up. In our case, the patient did well postoperatively and no recurrence was noted till date.

CONCLUSION

Myoepithelioma- plasmacytoid variant of the palatal - minor salivary glands is a rare entity. It is relatively more aggressive than other benign neoplasms of salivary glands. Management is surgical excision which should include margins of normal tissue and long term follow up for recurrence.

To conclude, myoepithelioma should be kept in mind as differential diagnosis when dealing with an intraoral sub mucosal mass inspite of their rarity at that location.

Conflict of Interest: Nil

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