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Plasmacytoid Myoepithelioma of the Hard Palate: A rare case Report

Medical Science

KEYWORDS : Myoepithelioma, minor salivary gland, hard palate

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ABSTRACT

Myoepithelioma is a benign tumor composed exclusively of neoplastic cells exhibiting myoepithelial differentiation. Myoepithelioma of salivary glands occur mainly in parotid gland accounting to 50%, followed by sublingual and submandibular glands. Myoepitheliomas of minor salivary glands accounts to only 1.0% of all salivary gland tumors. The peak age is from the third to fifth decade without any gender predominance. The vast majority of the intraoral minor salivary gland lesions are of palatal origin. Here we are reporting a case of a female of 25 yrs of age presenting with a mass in hard palate which histologically confirmed to be myoepithelioma of minor salivary gland in palate. Immunohistochemical analysis with S100 Pancytokeratin and Vimentin further supported the diagnosis.

Introduction

Myoepithelial cells are contractile flattened cells that surround both serous and mucous acini which, on contraction, force secretion from the acinar lumen into the duct system. They are located between the cell membrane of the secretory cells in acini and the surrounding basement membrane[1]. Normally, myoepithelial cells are essential component of some exocrine glands such as salivary glands, lacrimal glands, sweat glands and mammary glands[2].

Myoepithelioma of salivary glands occur mainly in parotid gland accounting to 50%, followed by sublingual and submandibular glands. Myoepitheliomas of minor salivary glands accounts to only 1.0% of all salivary gland tumors[3]. The tumors mostly present as asymptomatic, slowly progressive masses over a period of months to years in the patient with average age in the fourth decade with no sex predilection[3,4]. Myoepitheliomas of the minor glands are usually composed of plasmacytoid hyaline cells [2]. Here, we report a case of Plasmacytoid Myoepithelioma of palate focusing on the histological and immunohistochemical features. To our knowledge, there have been published only 24 previous such cases in the English literature.(Table 1)

Case	Age	Sex	Location	Treatment
Kahn and Schoub 1973	17y	F	Hard palate	Surgery
Sciubba and Goldstein 1976	22y	M	Right palate	Electrosurgery
Nesland et al. 1981	18y	F	Anterior soft palate	Surgery
Barnes et al. 1985	24y	F	Midline hard palate	Surgery
Lins and Gnepp 1986	8y	F	Right soft palate	Surgery
Ohtake et al. 1997	26y	F	Right palate	Surgery
Kanazawa et al. 1999	42y	F	Left hard palate	Surgery
Taylor and Tighe 1999	85y	F	Right soft palate	Surgery
Makoto et al. 2003	65y	F	Right palate	Enucleation
Bakshi et al. 2007 case 1	38y	M	Right soft palate	Surgery
Bakshi et al. 2007 case 2	35y	M	Left hard aplate	Surgery
Bakshi et al. 2007 case 3	65y	M	Right hard palate	Surgery
Zelaya et al. 2007	28y	F	Left hard palate	Surgery
Cruz Perez et al. 2007	13y	M	Hard and soft palate border	Surgery
Kim et al. 2007 case 1	63y	M	Posterior soft palate	Surgery
Kim et al. 2007 case 2	65y	F	Left soft palate	Surgery
Rastogi et al. 2008	33y	M	Hard palate	Surgery
Kim et al. 2009	52y	F	Left hard palate	Surgery
Zormpa et al 2010	29y	F	Hard and soft palate border	Surgery
Hakeem et al 2013	21y	F	hard palate	Surgery
Yadav et al 2013	40y	M	Right soft palate	Surgery
Swarna et al 2014	51y	F	Right hard palate	Surgery
Kulkarni et al 2015	11y	M	hard palate	Surgery
Chandra et al 2015	46y	F	hard palate	Surgery
Present case	25y	F	hard palate	Surgery

Table 1-Reported cases of myoepithelioma till date

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Case report

A 25 year old lady came to ENT department of RIMS, Ranchi, India with complains of swelling in right side of hard palate from 2 years. The swelling was gradually progressive and painless in nature. There was no history of dysphagia, odynophagia, voice change, loss of appetite, weight loss, bleeding from the lesion or fever. On examination there was a well-circumscribed, non-tender, non-pulsatile, firm, round, smooth surfaced pinkish mass located in right side opposite the upper right first molar tooth measuring 2.5×1.5×1cm^3 covered with normal mucosa. Regional lymph nodes were normal. Systemic examination of the patient was normal. A clinical diagnosis of benign palatal tumor was considered and a total excision of the lesion was carried out under local anesthesia The tissue was then sent for histopathological examination. The specimen was fixed in 10% buffered formaldehyde and embedded in paraffin. Serial sections (4 μm thickness) were taken from the block and stained with hematoxylin-eosin (H + E).

Grossly, the lesion consisted of fragments of gray-white tissue measuring 2×1.5×1cm^3. Microscopic examination revealed nests of tumour masses separated by sclerohyaline septa, numerous plasmacytoid cells with eosinophilic homogenous cytoplasm and eccentric round to ovoid vescicular nuclei .The mass was surrounded by a thin, fibrous capsule. There was no evidence of malignancy such as cellular pleomorphism, mitotic figures, necrosis, interstitial hemorrhage or infiltration of the adjacent tissues. (Fig.1,2.) The histopathological diagnosis was consistent with benign myoepithelioma of plasmacytoid type. Immunohistochemical studies were performed by avidin–biotin peroxidase complex technique. All stains were performed using commercially available Novocastra liquid mouse monoclonal antibodies. Immunohistochemical analysis revealed strong and diffuse positivity of all myoepithelial cells against Pancytokeratin, Vimentin and S-100 protein. GFAP immunostaining was negative.(Fig.3,4,5,6,7)

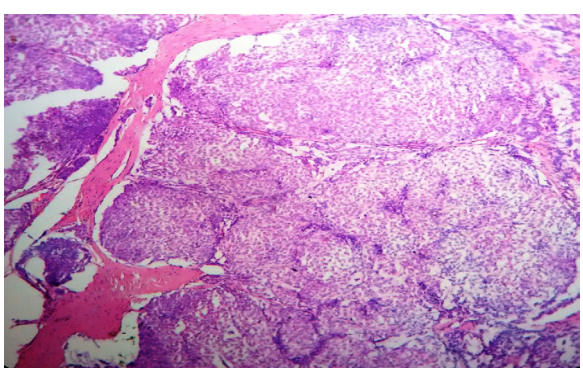

Fig.1-Nests of tumour masses separated by schlerohyaline septa(H+E stain 10X)

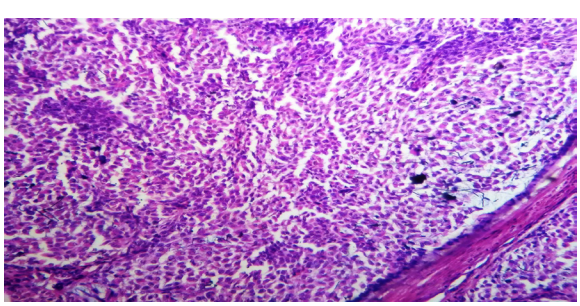

Fig.2-Section showing plasmacytoid hyaline cells with schlerohyaline septa in the right(H+E 20X)

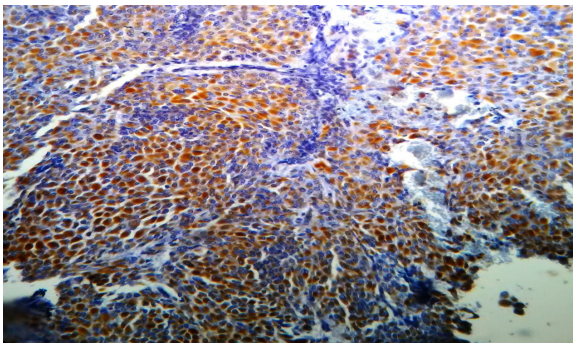

Fig.3-Diffuse cytoplasmic immunopositivity for pancytokeratin

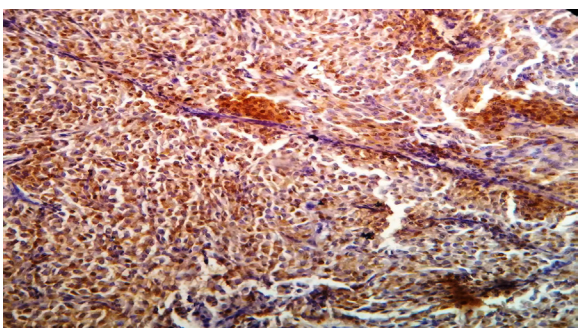

Fig.4-Diffuse cytoplasmic immunopositivity for S100

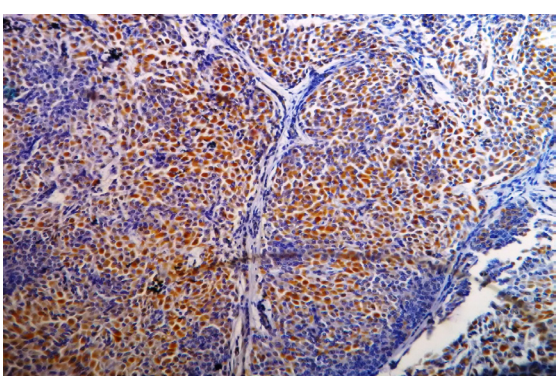

Fig.5-Diffuse cytoplasmic immunopositivity for Vimentin

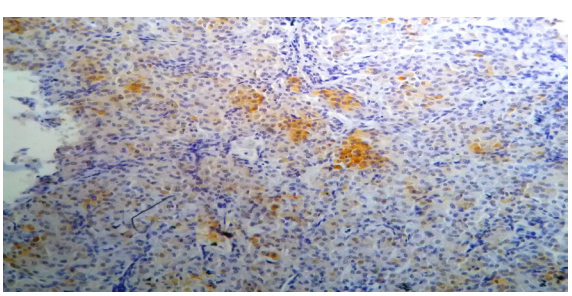

Fig.6-GFAP immunostaining was negative in this case.

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%)[5] . Myoepithelial cells are a component of most types of benign and malignant salivary gland tumors, particularly benign mixed

tumor, adenoid cystic carcinoma, and terminal duct carcinoma. Tumors thought to be composed exclusively of myoepithelial cells are generically referred to as myoepitheliomas. Myoepitheliomas frequently mistaken for cellular Pleomorphic Adenoma, because both the tumors consist of plasmacytoid cells[6]. The rarity of Myoepithelioma makes it a surprise diagnosis; identifiable only histopathologically. The neoplastic myoepithelial cells can be spindled, plasmacytoid hyaline, epithelioid, clear, or oncocytic, with the first two cell types being most common [2,4] .The hyaline (plasmacytoid) cell type tumors are composed of cells with eccentric nuclei, some degree of pleomorphism and hyperchromasia, but scanty or no mitotic activity. The cytoplasm is abundant, with a diffuse homogenous eosinophilia. Myoepitheliomas show four different morphological patterns which include nonmyxoid (solid), myxoid (pleomorphic adenoma like), reticular (canalicular like), and mixed[7]. Myoepithelioma of the palate clinically presents as slow growing, painless, firm submucosal mass as it was seen in our case. The differential diagnoses of intraoral submucosal mass include inflammatory abscess, deep mucocele, and salivary gland tumours. Plasmacytoid myoepithelioma needs to be differentiated from benign and malignant tumours like myoepithelial cell predominant pleomorphic adenoma, plasmacytoma, lymphoma, skeletal muscle and rhabdoid tumours[8]. Absence or less than 5% of epithelial cells showing ductal or acinar formation and absence of chondroid stroma helps in differentiating from pleomorphic adenoma. In our case no chondromyxoid stromal fragments were noted and inconspicuous ductal or acinar pattern helped us to distinguish it from pleomorphic adenoma. Absence of cellular pleomorphism, increased mitotic count and necrosis helps to differentiate myoepitheliomas from myoepithelial carcinomas[9]. Myoepitheliomas were considered as variant of pleomorphic adenoma; however, it is considered as separate clinical entity according to WHO since 1991 . In myoepitheliomas, the ducts constitute less than 5% of the section[10].

There are many immunohistochemical studies which demonstrate the variability of myoepithelial neoplastic cells' expression against some certain antibodies.Pancytokeratin,S-100 protein, CK(AE1/AE3), CK(5/6), vimentin and calponin are consistently positive, but the frequency of positivity and percentage of positive cells with the individual markers are highly variable[3]. S100 has been reported to be the most useful marker, but it lacks specificity[11]. For GFAP (Glial Fibrillary Acid Protein), tumor cells may exhibit either positive or negative reaction[2,12,13]. Ultrastructural studies can help confirm myoepithelial differentiation by identifying both epithelial (hemidesmosomes) and myoid features (myofilaments with focal densities, pinocytotic vesicles)[3].

Conclusion

Myoepithelioma of minor salivary glands are rare tumours and they must be considered in the differential diagnosis of any mass present in the hard palate. They must also be distinguished from other salivary glands tumors like pleomorphic adenoma and adenoid cystic carcinoma. Immunohistochemical analysis is a must for confirmation of this rare tumour.

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