Case Report

Myoepithelioma of Parotid: A diagnostic dilemma

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Abstract
Myoepitheliomas are rare benign salivary gland neoplasms which originate from ectodermally derived contractile smooth muscle cells i.e. myoepithelial cells which lack ductal differentiation. They account for only 1-1.5% of all salivary gland neoplasms. Myoepithelioma was considered as a variant of Pleomorphic adenoma in the past, but since 1991 it has been considered as a separate clinical entity by WHO. Their preoperative diagnosis on cytologic features is possible but is often difficult. Here we are discussing a case of parotid swelling which was cytologically diagnosed as basal cell adenoma but on histopathological and immunohistochemical studies, it was proved as a case of myoepithelioma.

Keywords: Myoepithelioma, Parotid, salivary gland

1. Introduction
Myoepitheliomas are rare benign tumors of salivary glands, accounting for only 1% - 1.5% of all salivary gland neoplasms. Less than 100 cases have been reported in the literature till 2013. The most common site of occurrence is parotid gland (50%) followed by sublingual gland (33%), submandibular gland (13%) and very rarely in oral cavity (1.5%). The varied histomorphology of these tumors, further confounded by their varied clinical behaviour presents many gray areas which need to be further studied. Although the cytological features are documented in a few case reports, it has rarely been diagnosed preoperatively by fine needle aspiration cytology.

2. Case report
A 45-year-old lady presented with complaint of swelling parotid region for past 2 years. On examination, swelling was 3 cms in diameter and firm in consistency. Fine Needle Aspiration was done and the smears showed round to ovoid cells arranged in clusters, trabeculae and groups as well as dispersed singly. These cells had round to ovoid nuclei with bland granular chromatin. Red to purple acellular hyaline matrix was seen as globules surrounded by tumour cells and as fibrillar substance. A cytological diagnosis of a benign neoplasm of salivary gland origin possibly basal cell adenoma was given. A superficial parotidectomy was done and tissue sent for histopathological diagnosis. Grossly the resected salivary gland tissue showed a well circumscribed grey white to grey brown area measuring 3 cms in diameter. The histopathological examination showed an encapsulated lesion composed of round to plasmacytoid and clear cells forming trabeculae, cords and acinar formations with interspersed hyaline material. The ductal component was not evident in the histological sections. There was no evidence of necrosis or abnormal mitotic activity. Immunohistochemically, the tumour cells showed positivity for S100 and focal positivity for SMA. So a final histological diagnosis of Myoepithelioma was given.

Figure 1 – Leishman stained cytological smears (100 X) showing tumour cells arranged in groups, trabeculae and acini with acellular hyaline material in between.

Figure 2 – Leishman stained cytological smears (400 X) showing round to ovoid tumour cells forming acini around globules of hyaline acellular material.
3. Discussion

Myoepitheliomas are rare benign salivary gland neoplasms. They originate from ectodermally derived contractile smooth muscle cells i.e. myoepithelial cells which lack ductal differentiation. Hence, it is reported at sites where myoepithelial cells are present e.g. in salivary glands, breast, larynx & sweat glands of skin. Myoepithelial cells are most commonly present in parotid gland, thus tumour is also most common in this gland. In our case too tumour was seen involving the parotid gland. The tumour usually presents as a slowly growing, painless, asymptomatic mass in fourth decade of life. The patient in this case also was a 45-year-old lady. However other studies show no predilection with regard to gender and age.

Myoepitheliomas were considered as a variant of Pleomorphic adenoma in past, but since 1991 it has been considered as a separate clinical entity by WHO. Myoepitheliomas of parotid usually present as encapsulated lesions with no ulceration. Pleomorphic adenoma, basal cell adenoma and myoepithelioma are benign salivary gland tumors with myoepithelial cell participation that appear to lie in a continuum. Histologically, myoepithelioma can be viewed as an extreme form of basal cell adenoma without a ductal component (less than 5-10%) while basal cell adenoma is pleomorphic adenoma minus the characteristic stroma. A definite diagnosis and differentiation between these remains difficult on Fine needle aspiration cytology. In present case too, initial cytological diagnosis diagnosis given was of a benign salivary gland neoplasm possibly basal cell adenoma.

Myoepitheliomas show four different morphological cellular patterns that consist of mainly spindle cell, plasmacytoid, clear cell and oncocytic (a variant of spindle cells) types. To consider a diagnosis of pure myoepithelioma, the epithelial component should be less than 5% - 10% and fibromyxoid stroma should be absent. Identification of myoepithelioma is important as WHO classification attributes a more aggressive behaviour to it as compared to Pleomorphic adenoma.

In some studies possibility of malignancy arising in myoepithelioma is 20% as compared to 5.2% in Pleomorphic adenoma. Also, differentiation of myoepithelioma from its malignant counterpart i.e. malignant myoepithelioma is also crucial as it has a more aggressive clinical behaviour, may recur even after adequate treatment or metastasize. Histopathologically, presence of cellular atypia, cellular pleomorphism, necrosis, increased mitotic figures and infiltrative growth pattern favour the diagnosis of malignant myoepithelioma. Myoepitheliomas show variable results on immunostaining. They generally show positive or focally positive staining for pancytokeratin, CK-14, S100, GFAP, calponin and actin. In this particular case also, S100 and SMA were positive.

The treatment of choice of myoepithelioma is surgical excision with a margin of uninvolved tissue around. Incomplete resection may lead to recurrence in 15% - 18% of cases.

4. Conclusion

Myoepithelioma is a rare salivary gland tumour which may arise in unusual location and awareness about its varied morphological pattern augmented by immunohistochemistry helps in its identification. This is important as myoepitheliomas may follow an aggressive behaviour and most of them can be cured by adequate excision. Their pre-operative diagnosis on cytologic features is possible but is often difficult. Nevertheless, cytologic features together with immunocytochemical studies can be utilised for a definite diagnosis on FNAC in suspected cases preoperatively to help plan the treatment protocol & extent of surgery.

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