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Case Report

Uncommon plasmacytoid myoepithelioma originating from ectopic minor salivary gland in neck: a rare case with rare site presentation

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ABSTRACT

The aim of this case report is to present as case of plasmacytoid at ectopic minor salivary gland in the neck which is a rare neoplasm with uncommon presentation of site. Usually it is a tumor of major salivary gland and more common in parotid gland and less common in the minor salivary gland of the oral cavity predominantly in palate. Here authors are discussing a case of 10-year-old female who presented in our institute with complain of swelling over right mid cervical region which was clinically suspected as reactive cervical lymph node. Patient was evaluated further and cytologically diagnosed as a case of plasmacytoid myoepithelioma. It was also confirmed as a case of plasmacytoid myoepithelioma after histopathology and immunohistochemistry studies. Plasmacytoid myoepithelioma at ectopic minor salivary gland site in the neck is uncommon presentation and only a limited number of cases have been reported in literature.

Keywords: Plasmacytoid, Minor salivary gland, Neck

INTRODUCTION

Sheldon is the first who described the entity of myoepithelioma in 1943. Previously it was considered as a variant of pleomorphic adenoma but who, in 1991 considered as a distinct entity based on histopathological features.[1]

Salivary glands tumour accounts for up to 6.5% of all head and neck neoplasms. Among these myoepithelioma represents approximately 1-1.5% of salivary gland tumour. Nearly 40% arise in the parotid gland. Myoepithelioma is the rare benign tumour of the head and neck. The growth pattern is solid, myxoid or reticular, composed may be clear cell type, spindle shaped, plasmacytoid and epithelioid.[2] There is no sex predilection and the peak age of occurrence is in third decade of the life. When benign, pain less swelling is the most common symptom with the duration of ranging from 2 months to 7 years.

Confirmatory diagnosis of myoepithelioma need histo-pathology, immunohistochemistry that typically show positivity for cytokeratin and s-100, calponin, smooth muscle actin myosin vimentin, glial fibrillary acidic protein and carcinoembryonic antigen but the main markers for myoepithelioma is the s-100 protein.[3]

CASE REPORT

A 10 years old female child presented for evaluation of a firm to hard palpable nodular swelling on the right mid cervical region of neck. The nodular had developed since 6 months previously with negative history of increases in size. On examination, there was firm to hard, non-tender and mobile swelling. The swelling was 0.8×0.8 cm in dimension. The skin overlying the swelling was free. Mass was clinically initially suspected as reactive lymph node. Fine needle aspiration (FNAC) cytomorphology of the swelling was done under aseptic precaution, using a half inch 22-inch gauge needle. Smears were prepared and analysed microscopically, after stained with Papanicolaou and Leishman stain. Revealed discrete and cohesive cluster of mononuclear cells showed plasmacytoid nuclear appearance and moderate amount eosinophilia cytoplasm.

Few cells entrapped in myxoid material. Mitosis and cellular pleomorphic were seen. The mass was easily excised surgically and fixed 10% formalin

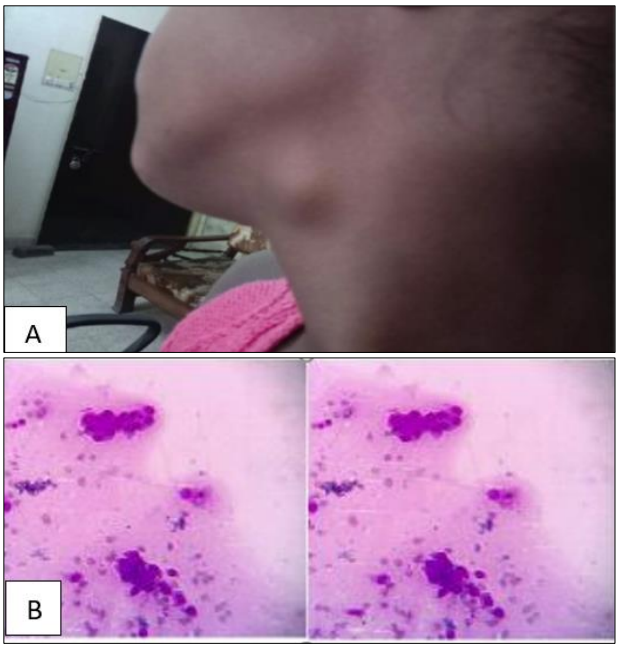

Figure 1: (A) Leishman stained cytology smears of tumor cells arranged in groups. (B) Leishman stained cytology smears of tumor cells arranged in groups and scattered having round to ovoid eccentric nuclei (400×) and gross picture of grayish-white to grayish-brown solid tumor mass scattered having round to ovoid eccentric nuclei (400×).

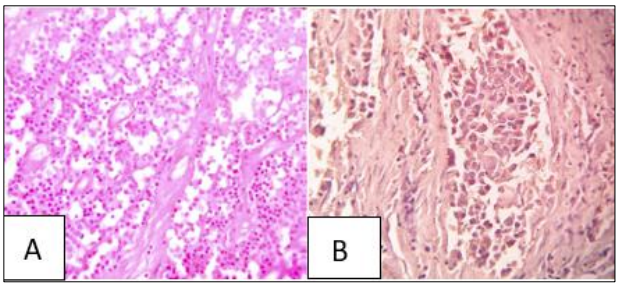

Figure 2: (A) Histology micrograph showing uniform round to plasmacytoid tumour cells separated by collagen stroma (H and E; 100×). (B) Immunohistochemical staining, S100 positivity (100×).

Grossly, received specimen measured 1.0×0.08×0.05 cm in diameters. The tumour was greyish white to greyish brown in colour. Tissue was processed, sectioning was done and stained with haematoxylin and eosin (H and E). Histopathological examination showed tumour surrounded by thin fibrous capsule, composed of round to tissue, had eccentrically located nuclei and an abundant, dense, hyaline eosinophilic cytoplasm without nuclear atypia or mitoses. Tumour cells showed immunoregulative for epithelial membrane antigen. Finally, the case was diagnosed as plasmacytoid myoepithelioma after histopathological examination (HPE) and immunohistochemistry (IHC).

DISCUSSION

Myoepithelioma is disease of myoepithelioma differentiation with unknown aetiology which reportedly behaves in a benign manner. It is the global in distribution and affects all ages ranging 9-85% with an incidence peak in the thirties with equal frequency in both male and female. The commonest presentation is as asymptomatic and slowly, growing mass, the presentation of tumour depends upon the location. Observation of site involvement will done most common location of this tumour in the head and neck is parotid gland and or accessory glands of the oral cavity in the present study, the location was left mid cervical of neck.[4] It is very difficult to diagnose clinically since it is an unusual site, hence, most the patients are diagnosed after attempting FNAC. After searched many literatures we had found that palate affair is the common. Other sites like orbit. Breast tongue intraosseous, maxilla, kidney and ear were also involved. Myoepithelioma have four different morphological pattern dwells of mainly spindle cell. Plasmacytoid, clear cell and oncocytic variant of spindle cells type and also and also display variable growth patterns like non-myxoid, myxoid, reticular and mixed.[5] To consider a diagnosis of pure myoepithelioma the epithelial component should be absent or less than 5-10% and fibro myxoid stroma should be absent or less. Yoshihiro et al in their study said that distinguishing RRC and malignancies of salivary gland origin is very important.[6] They presented a case which had history of RCC and myoepithelioma. Metastasis from RCC most commonly affects the lungs, bone liver, adrenal glands, contralateral kidney, and brain. When it was diagnosed first considered a variant of pleomorphic adenoma but after 50 year later it was described distinct entity. A case study of 38 patients were done by Vickie et al sign of lesions ranged from 0.3 to 2.7 cm in size with the patient's age ranged between 2 months to 74 years.[7] The anatomic distribution of ase was as follows: upper extremities, shoulder, lower extremities back face chest and buttock.

Histology of myoepithelioma of the hard a plate showed cluster and sheets of homogenous plasmacytoid cells have round or ovoid eccentric nuclei. Plump spindle cells in myxoid background of FNAC of the thyroid nodule. Immunohistochemistry helps in its identification. Final conclusion is that myoepithelioma is a benign tumour of myoepithelial origin.

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