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RESEARCH ARTICLE

NARRATION OF UNCOMMON PLASMACYTOID MYOEPITHELIOMA ORIGINATING FROM RIGHT NECK REGION: AN EXTREMELY RARE CASE WITH REVIEW OF LITERATURE

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ABSTRACT

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

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INTRODUCTION

Sheldon is the first who described the entity myoepithelioma in 1943. Previously it was considered as a variant of pleomorphic adenoma but World Health Organization, in 1991 considered it as a distinct entity on the basis of histopathological features (Mehta et al., 2017; Vinicius et al., 2017; Vani et al., 2014). Salivary gland tumors account for up to 6.5% of all head and neck neoplasms. Among these myoepithelioma represents approximately 1-1.5% of salivary gland tumors. Nearly 40% arise in the parotid glands (Richa et al., 2012; Daspin et al., 2016). Myoepithelioma is the rare benign tumor of the head and neck. The growth patterns may be solid, myxoid or reticular, composed of sheets and islands of myoepithelial cells and component may be clear-cell type, spindle-shaped, plasmacytoid and epithelioid. There is no sex predilection, and the peak age of occurrence is in the third decade of life (range= 9 - 85 yr) (Ferri et al., 2006; Saurabh et al., 2013). When benign, painless swelling is the most common symptom with the duration of ranging from 2 months to 7 years. Other complains are related to the involved site (Yousif et al., 2017).

Confirmatory diagnosis of myoepithelioma need histopathology and Immunohistochemistry that typically show positivity for cytokeratin and S-100, calponin, smooth muscle actin, myosin, vimentin, glial fibrillary acidic protein (GFAP), and carcinoembryonic antigen but the main marker for myoepithelioma is the S-100 protein (Gustavo et al., 2017).

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 swelling had developed since 6 months previously with negative history of increase in size. On examination, there was a firm to hard, non-tender, and mobile swelling. The swelling was 0.8X0.8 cm in dimension. The skin overlying the swelling was free. Mass was clinically initially suspected as reactive lymph node. Fine needle aspiration cytology (FNAC) of the swelling was done under aseptic precaution, using a half inch, 22-gauge needle. Smears were prepared and analyzed microscopically after stained with Papanicolaou and Leishman stains, revealed discrete and cohesive clusters of mononuclear cells showed plasmacytoid nuclear appearances and moderate amount of eosinophilic cytoplasm. Few cells entrapped in myxoid material. Mitosis and cellular pleomorphism were absent with no ductal and acinar cell groups were seen. (Image1-a) The mass was easily excised surgically and fixed in 10% formalin.

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Grossly, the postoperative specimen measured 1.0X 0.8x0.5 cm in diameters. The tumor was solid, grayish-white to brown in color (Image1-b). Tissue was processed, sectioning was done and stained with hematoxylin and eosin (H & E). Histopathological examination (HPE) showed tumor surrounded by thin fibrous capsule, composed of round-to-polygonal shaped cells intermixed in a stroma of dense fibrous tissue, had eccentrically located nuclei and an abundant, dense, hyaline, eosinophilic cytoplasm without nuclear atypia or mitoses. Tumour cells showed immunopositive for the expression of S-100 and immunonegative for epithelial membrane antigen (Image 2-a & b). Finally, the case was diagnosed as plasmacytoid myoepithelioma after HPE and IHC.

DISCUSSION AND REVIEW OF LITERATURE

Myoepithelioma is a disease of myoepithelial differentiation with unknown etiology which reportedly behaves in a benign manner.

It is global in distribution and affects all ages, ranging 9-85 with an incidence peak in the 30s, with equal frequency in both males and females. Kim *et al* reported a case of cutaneous myoepithelioma neck in 53 years old women. Similar case was also found reported by Mardi *et al* in 25 years old male. In present case was a 10 years old young female patient. Other than these two cases, we have not found so much previous report of this tumor occurring in the neck and the result said, this benign tumor could be presented at any age and sex (Saurabh *et al.*, 2013; Kim *et al.*, 2016; Mardi, 2007).

The commonest presentation is as asymptomatic and slowly growing mass. The presentation of tumor depends upon the location (Politi *et al.*, 2005). Spinal cord myoepithelioma causes weakness of both lower limb, bowel and bladder incontinence in the study of Daspin *et al.* Mode of presentation is varied in patients with other studies done by Mehta K and Louis MB etc (Mehta *et al.*, 2017; Daspin *et al.*, 2016; Louis *et al.*, 2018).

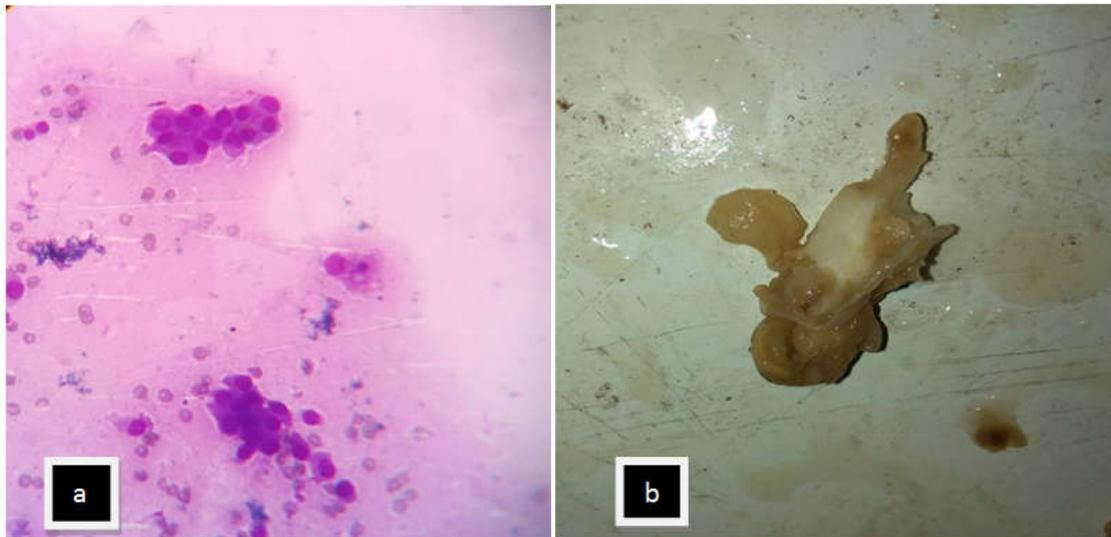


Image 1.(a) Leishman stained cytology smears showing tumor cells arranged in groups and scattered having round to ovoid eccentric nuclei (400 X) (b) Gross picture of grayish-white to grayish-brown solid tumor mass

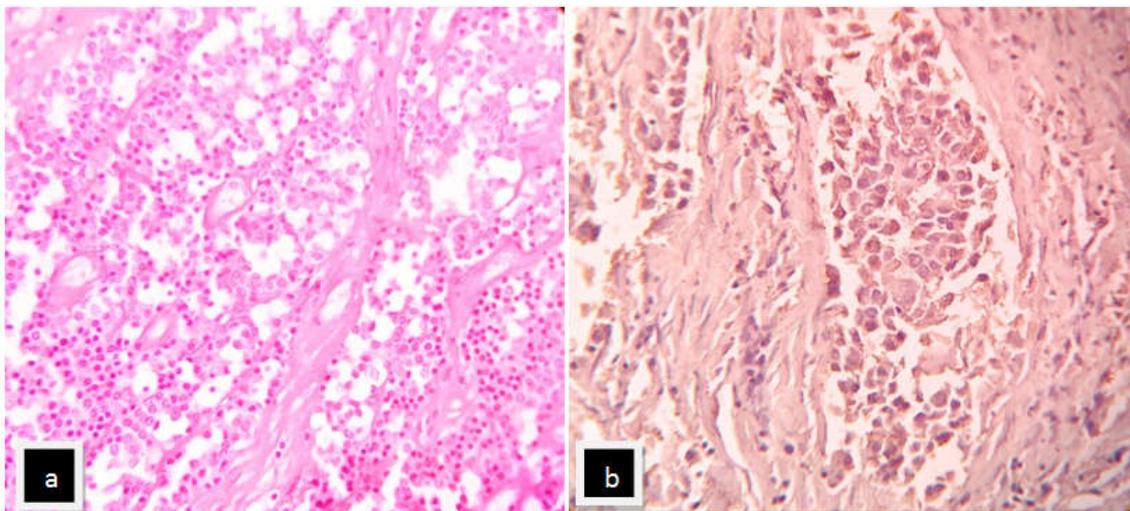


Image-2: (a) Histology micrograph showing uniform round to plasmacytoid tumour cells separated by collagen stroma (H&E; 100X) (b) Immunohistochemical staining, S100 positivity (100X)

In our patient presenting feature was palpable mass. In CT scan, myoepithelial carcinoma appears isodense to muscle and shows moderate homogenous enhancement with contrast. On MRI, it is hypointense on T1 and shows a dense homogenous enhancement (Saha *et al.*, 2017). In the present case report, we cannot comment of CT and MRI findings as the reports were lost by the patient. Before analyzing cellular pattern and morphology elements; observation of site involvement will be done. Most common location of this tumor 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 (rarest site). It is very difficult to diagnose clinically since it is an unusual site. Hence, most the patients are diagnosed after attempting FNAC. Many publications defined its different location. Myoepithelioma can occur many sites as mentioned in literature. Swati and Teruo Shiomori reported a case myoepithelial carcinoma in in the upper lip (unusual location) (Saurabh *et al.*, 2013; Gotmare *et al.*, 2017; Teruo *et al.*, 2005). Myoepithelial carcinoma with myoepithelioma occurring in the right forearm was diagnosed in the case report of Youssef Mahdi *et al.* (2014) After searched many literature we had found that Palate affair is the common after parotid gland also in hard palate is common. Other sites like orbit, breast, tongue, intraosseous, maxilla, kidney and ear were also involved (Kapila *et al.*, 2011; Khan *et al.*, 2013; Deepthi *et al.*, 2015; Bharat *et al.*, 2014; Sheeraz *et al.*, 2013; Feng *et al.*, 2015; Vedula *et al.*, 2014).

Myoepithelioma have four different morphological pattern dwell of mainly spindle cell, plasmacytoid, clear cell and oncocytic variant of spindle cells types and also can also display variable growth patterns like non-myxoid(solid), myxoid (pleomorphic adenoma-like), reticular (canalicular-like) and mixed. To consider a diagnosis of pure myoepithelioma, the epithelial component should be less than 5% - 10% and fibromyxoid stroma should be absent or less (Puja *et al.*, 2014; Anmolsingh *et al.*, 2017). Yoshihiro *et al* in their study said that distinguishing RCC and malignancies of salivary gland origin is very important. They presented a case which had history of RCC (26 years back) and myoepithelioma (7 years back). Metastasis from RCC most commonly affects the lungs, bone, liver, adrenal glands, contralateral kidney, and brain (Yoshihiro *et al.*, 2018). Diagnosing a neck nodule remains a challenge for the clinician since many distinct entities present with the same clinical features. Among them myoepithelioma is a rare tumor at this site. As I have mentioned previously that I did not find so many cases. When it was diagnosed first, considered as a variant of pleomorphic adenoma, but after 50 years later it was described distinct entity. A case study of 38 patients were done by Vickie *et al.* (1997-2012) sign of lesions ranged from 0.3 to 2.7cm in size with the patient's age ranged between 2 months to 74 years. The anatomic distribution of cases was as follows: upper extremity (11), shoulder (3), lower extremity (15), back (6), face (2), chest (1), and buttock (1). None had been described on the neck. Study of 101 cases by Hornick *et al.* described that majority arose in the lower limb/limb girdles followed by upper limb/limb girdles, head, neck and trunk. Only 5 (5.0%) tumors were located in the buttock area (Kim *et al.*, 2016; Louis *et al.*, 2018; Bhanvadia *et al.*, 2017). Myoepithelioma is a rare benign tumor but after review many literature I realized that this is not so rare as mentioned in many books and manuscripts. Sethi *et al* described histology of myoepithelioma of the hard palate showed clusters and sheets of homogenous plasmacytoid cells have round or ovoid

eccentric nuclei (Sethi *et al.*, 2012). Narick *et al.* described plump spindle cells in myxoid background on FNAC of thyroid nodule. Harada *et al.* described myoepithelioma in the forearm showing tumor cells were arranged in a reticular and/or trabecular fashion with a myxoid background. Rekhi *et al.* described a case of Intraosseous myoepithelioma in the left leg (Narick *et al.*, 2015; Harada *et al.*, 2015; Rekhi *et al.*, 2014). Final conclusion is that Myoepithelioma is a benign tumor of myoepithelial origin but not a rare as much. It can arise at many unusual locations. We should aware for its varied origin and morphology. Immunohistochemistry helps in its identification.

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