Myoepithelioma of the Lateral Border of Tongue - A Rare Site of Occurrence and Dilemma of Diagnosis

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PRESENTATION OF CASE

A 62 year old female patient reported with a chief complaint of swelling in the tongue (Figure 1) from the past six months. The swelling was painless, and gradually grew in size. On examination the mass was roughly 3 cm anterio-posteriorly and 3 cm in thickness. Firm on palpation and had definite border. Computer tomography conducted showed, a smooth well circumscribed ovoid mass located in the left lateral border of the tongue at around the middle third measuring about 3 cms in all directions (Figure 2).

CLINICAL DIAGNOSIS

Macroscopically the lesion appeared as well circumscribed and well encapsulated. Based on clinical scenario we suspected the lesion primarily to be a pleomorphic adenoma. As pleomorphic adenomas are the most common types of lesions on the tongue.

DIFFERENTIAL DIAGNOSIS

1. Granular cell tumour.
2. Basal cell Adenoma.
3. Neurofibroma.
4. Schwannoma.
5. Perineurioma.

PATHOLOGICAL DISCUSSION

Histopathological evaluation revealed spindle shaped cells proliferating in the form of sheets and interlacing fascicles. Little intercellular fibrous stroma was seen (Figure 3). The spindle cells appeared elongated with faintly eosinophilic cytoplasm and pale centrally placed nucleus. Mitotic activity and cellular pleomorphism was minimal.
Immunohistochemical analysis was done using p63, vimentin, SMA, S100, cytokeratin and MIB (Ki 67) was done. P63, vimentin, cytokeratin showed strong positivity (Figure 4). Cytokeratin stain showing strong positivity. SMA, S100 showed focal positivity (Figure 5).

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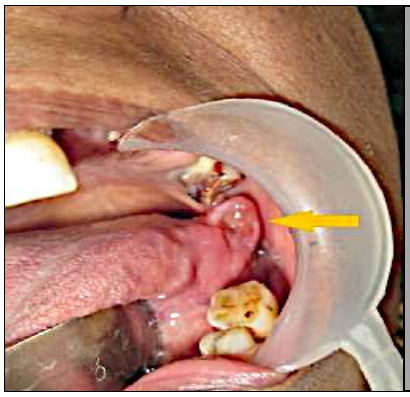

Figure 1.
Growth on the Lateral Border of the Tongue

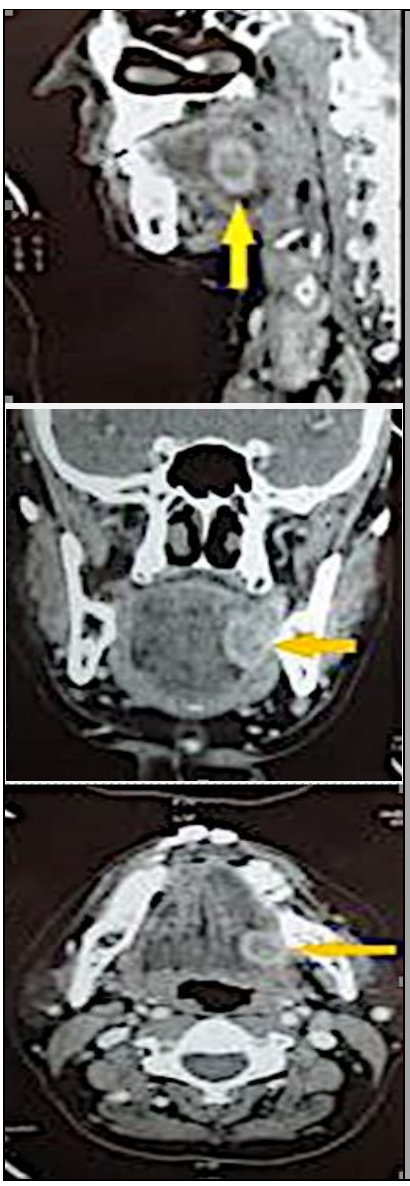

Figure 2.
CT Scan in Sagittal, Axial and Coronal Sections Showing a Well Circumscribed Encapsulated Oval Lesion with Well Demarcated Borders

Stains MIB or Ki67 was negative. By correlating the clinical, histological and immunohistochemical analysis a diagnosis of myoepithelioma was given. Myoepithelial neoplasms are tumours which are composed mostly of cells with myoepithelial differentiation behaving benignly and termed as myoepitheliomas. Incidence of myoepithelial neoplasm is about 1.5 % of all salivary tumours.[1-4] These tumours are commonly observed in adults aged between 30 and 80 years (average age 36.3 years).[5]

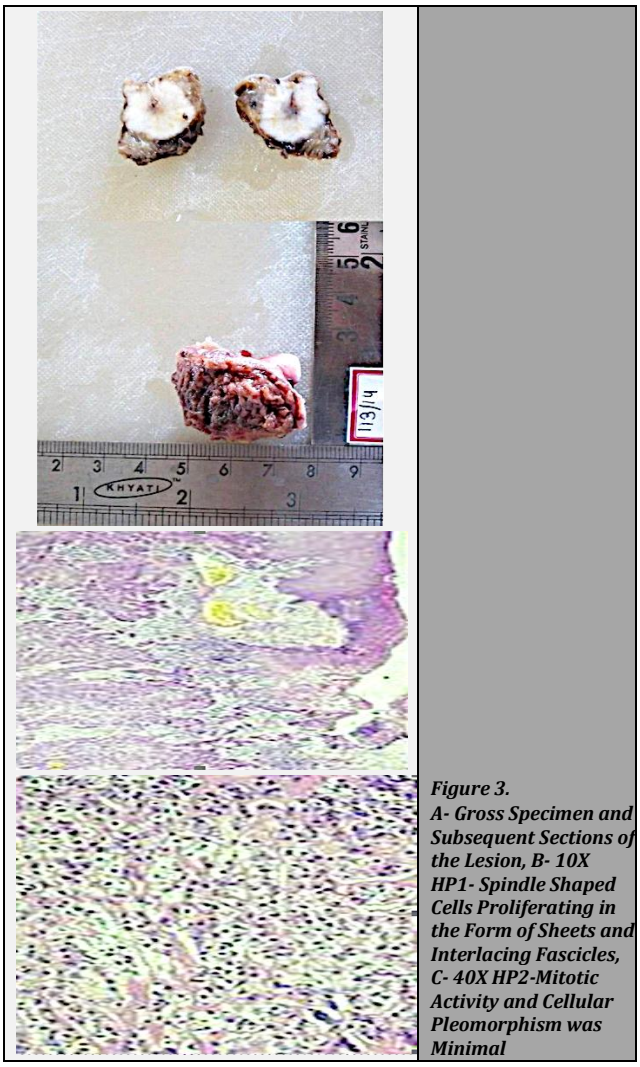

Figure 3.
A- Gross Specimen and Subsequent Sections of the Lesion, B- 10X HP1- Spindle Shaped Cells Proliferating in the Form of Sheets and Interlacing Fascicles, C- 40X HP2-Mitotic Activity and Cellular Pleomorphism was Minimal

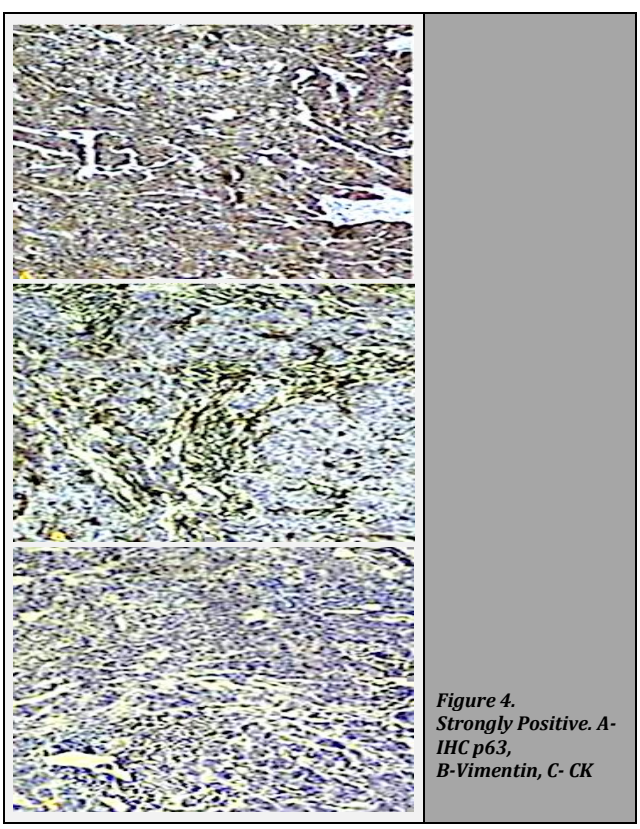

Figure 4.
Strongly Positive. A- IHC p63, B-Vimentin, C- CK

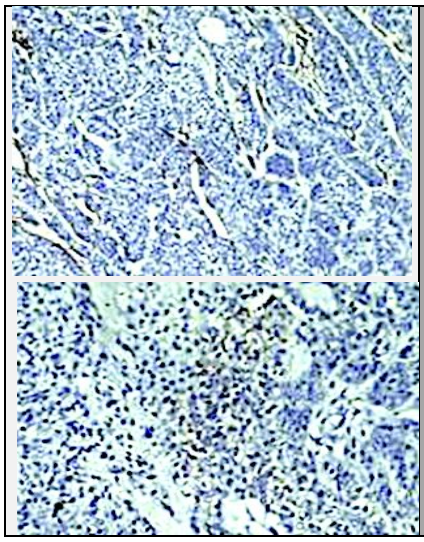

Figure 5.
Focal Positive IHC-SMA & S100

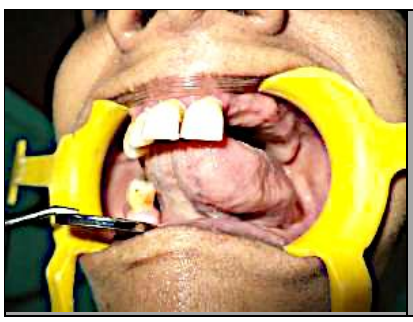

Figure 6. 7th Year Follow-Up, Disease Free State.

Myoepitheliomas has a diverse morphologic and immune-phenotypic variation, therefore cause difficulties in their diagnosis. Due such difficulties these tumours may be under-recognized and might not be as rare as reported.

Routine history taking and initial investigations like CT scans and FNAC (Fine Needle Aspiration Cytology) may not diagnose myoepithelioma at the earliest. This condition may be missed in investigations like FNAC because the cytological features of the disease are diverse,[3] they include spindled, stellate, epithelioid[1,5] plasmacytoid, basaloid, oncocytic, or clear cells.[1-4] It may be composed of spindle cells, which are arranged in interlacing fascicles that show stroma like appearance;[5] plasmacytoid cells that are polygonal cells with eccentric nuclei; or epithelial cells arranged in nests or cords. Various architectural patterns of arrangement of these cells are non-myxoid (solid), myxoid (pleomorphic adenoma like), reticular and mixed.[6]

Differentiation of myoepithelioma from other tumours, especially from pleomorphic adenoma may be potentially difficult owing to its varied architectural presentations. Myoepithelioma and pleomorphic adenoma have been suggested to be two different forms of the same entity.[7] Any neoplasm containing less than 5 % of ductal and acinar component is to be considered as myoepithelioma.[5]

Immunohistochemistry plays an essential role in identification of myoepithelial cells with reactivity for CKs and at least one of the other myoepithelial markers, which may include S100, vimentin, calponin, p63, glial fibrillary acidic protein, CD10, smooth muscle actin, and smooth muscle myosin heavy chains, is required for diagnosis[1]. The immunophenotype of each case is highly variable. Hence a wide spectrum of markers typical of myoepithelial immune profile has to be done.

Myoepithelioma is part of salivary gland tumours which includes pleomorphic adenoma and basal cell adenomas.[8] Other type of tumours in possible differential diagnosis includes soft tissues tumours such as leiomyoma, which are S-100 protein negative. Even though Schwannomas are S-100 positive, they tend certain typical microscopic features. Myoepitheliomas having Verocay bodies have been reported in literature.[9,10] Myoepithelial differentiation is seen in polymorphous low-grade adenocarcinoma in immunohistochemical stains and haematoxylin eosin sections and shows infiltrative nature unlike myoepithelioma which is usually well-circumscribed.[11]

Malignant tumours and their benign counterparts can be distinguished by their characteristic multi-lobulated architecture, presence of infiltrating growth, necrotic areas, polymorphism and mitotic figures.[12] A look into prevailing literature suggests that assessing the cell proliferative activity can hold the key to distinguishing between benign and malignant myoepithelioma. Also, Ki-67 labelling index of more than 10 % is diagnostic of myoepithelial carcinoma.[13,14,15,16] The present case on histopathological examination did not show any of these features and immune marker of proliferating cells Ki-67 was negative based on these findings, it was considered as a typical benign neoplasm.

DISCUSSION OF MANAGEMENT

Results of FNAC performed suggested basal cell adenoma. Subsequently the patient underwent complete excision of the lesion with 1 cm healthy margin of tissue. The entire specimen (Figure 3) was sent for histopathological examination. The wound was closed primarily. The patient has been on regular follow up and is disease free till date.

DISCUSSION

In head and neck neoplasm's salivary gland tumours constitute about 3 – 4 % of all types of tumours reported. Of these salivary gland tumours, nearly 80 % originate in the parotid gland, the other major salivary glands such as the submandibular gland or sublingual gland constitute to a much lesser extent.[1] Myoepithelioma is a rare tumour which mainly involves the parotid gland and has hardly been reported in the tongue. Statistically myoepithelioma accounts for less than 1 % of all major and minor salivary gland tumours. This tumour described by Sheldon in 1943 was later in 1991 named by the World Health Organization as pathologically distinct. Highly varied structural presentation of these tumours and associated difficulty in diagnosis paved the way for a new classification in 2005. A fundamental characteristic feature of myoepithelioma consists of myoepithelial cells with various growth patterns and appearances.[2]

We came to a final diagnosis of myoepithelioma a rare salivary gland neoplasm, after applying a panel of IHC (Immunohistochemistry) markers. Careful analysis is required from tumors arising from salivary glands as this group of tumours have a diverse nature and can vary in cell type and in immuno-histo-chemistry. The diagnosis requires confirmation by immunohistochemical analysis. Complete surgical excision with margin placed in healthy tissue area is

the treatment of choice with the recurrence risk being very low after removal.

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