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Myoepithelioma Of Parotid Gland: A Case Report

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Abstract

Salivary gland tumors are not encountered frequently. They usually have different histo-pathological subtypes. The overall incidence of salivary gland tumors is 2.5–3 per 100,000 patients per year. Among the various histological patterns and types, myoepithelioma of the parotid gland is very rare. Incidence of myoepithelioma is only 1–1.5% among all salivary gland tumors. Myoepithelioma is a benign salivary gland neoplasm formed almost entirely of myoepithelial cells arranged in a sheet, island or cord-like fashion. On CT scan, it presents as a well-circumscribed homogenous lesion with lobulated or smooth margins. Management of these tumors include complete surgical excision with wide margins. It has a very low recurrence rate compared to that of pleomorphic adenomas. Immunohistochemistry is of utmost importance in suspected myoepithelioma cases to ensure proper treatment and follow-up.

Herein, we describe a case of myoepithelioma in a 31-year-old male patient, who presented with a slow growing mass in the right parotid region. There were no aggravating and/or relieving factors found. On examination a 3 × 3 cm diffuse swelling was found below the right ear lobule. Fine Needle Aspiration Cytology (FNAC) was performed. The findings were suggestive of salivary gland tumor of low malignant potential. The mass was surgically excised for a definitive diagnosis. The final histopathology report along with immunohistochemistry revealed the tumor to be myoepithelioma of parotid gland.

Keywords: Parotid swelling, FNAC, IHC, histopathology, myoepithelioma, smooth muscle tumor

Introduction

Myoepitheliomas (MEs) are rare tumors of the parotid gland. Myoepitheliomas are generally seen in the major and minor salivary glands. Involvement of parotid gland is the most common, seen in approximately 40% of cases. Only 1% of all salivary gland neoplasms are myoepitheliomas. Myoepithelioma is usually a benign tumor arising from neoplastic myoepithelial which are found between the basement membrane and the basal plasma membrane of acinar cells. Majority of myoepitheliomas are benign but malignant transformation can take place in recurrent cases and also in cases which are left untreated.

Complete surgical excision is the routine management and follow up is needed to a check for any recurrences. Definitive diagnosis is made by extensive histopathological examination along with immunohistochemical work up. We present a case of myoepithelioma in an adult male patient in right parotid gland. In our case, a final diagnosis was given based on histology and IHC results.

Case Report

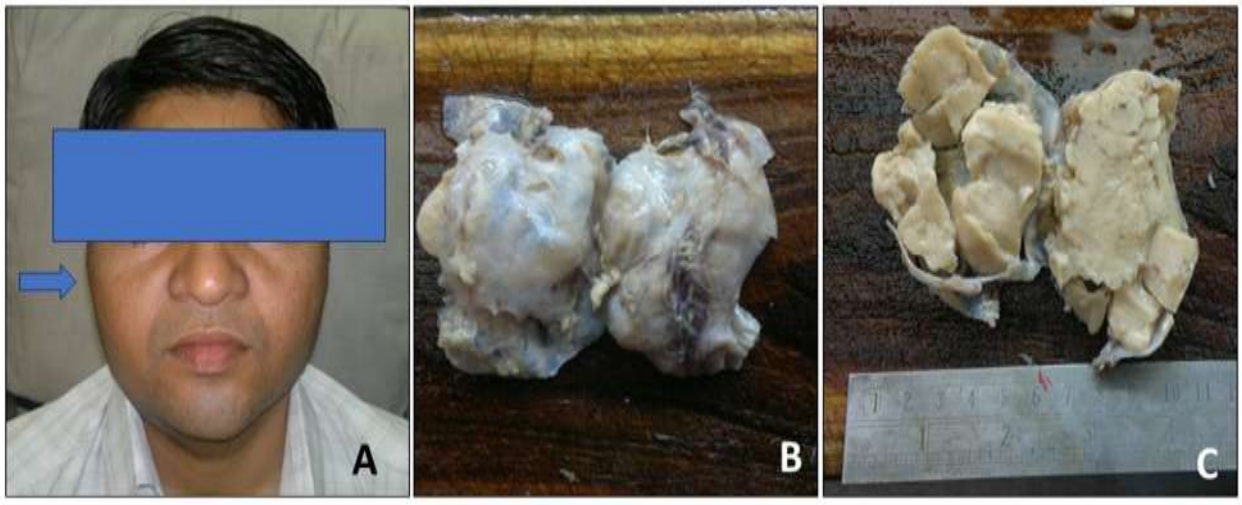
A 31-year-old male patient presented to ENT out-patient department with chief complaints of swelling in right parotid region for the last 6 months. The swelling was mobile and slowly progressive in nature, with no pain. On examination a diffuse swelling was noted just below the right ear lobule, with ill-defined borders [Figure 1A]. The swelling extended from angle of mandible to about 5 cm posteriorly and also from inferior wall of external auditory canal to 5 cm downwards. The swelling was slightly fixed and non-tender. The overlying skin was normal. On oral examination, a bulge was noted in the right parapharyngeal wall with uvula in the midline. No other swelling or any abnormality was noted on examination.

A provisional diagnosis of a parapharyngeal tumor was made on clinical grounds. The patient was sent for fine needle aspiration cytology (FNAC). Based on cytology, a diagnosis of Salivary gland tumor of low malignant potential was made. However, possibility of polymorphous low-grade adenocarcinoma could not be ruled out. Biopsy or surgical excision with detailed histopathological examination & immunohistochemistry was advised to arrive at a definitive diagnosis.

Contrast enhanced computed tomography (CECT) was done and showed a 7x4 cms, heterogeneously enhancing lesion in right pre-styloid parapharyngeal region involving the deep lobe of parotid. Widening of stylo-mandibular tunnel was noted. On radiology, a diagnosis of a parotid tumor (suspicious for malignancy) was made along with presence of 2, level 2 lymph nodes. Hence to rule out malignancy, a histopathological correlation was advised.

Through transcervical approach, under general anesthesia, the mass was surgically excised and sent for histopathological evaluation. On gross, a solitary, well encapsulated mass was received, measuring 3x3 cms [Figure 1B]. External surface was unremarkable. Cut sections showed grey-white areas [Figure 1C].

Fig1: A- Clinical image showing swelling below right ear lobule; B & C- Gross images showing grey-white encapsulated mass, cut section of ehich showed grey-white solid areas.



H&E-stained sections showed uniform round to oval cells arranged in sheets with bland chromatin in a hyalinized scant stroma [Figure2]. No necrosis, infiltration or any glandular pattern was noted in the sections examined. Based on histomorphology, the differentials considered were paraganglioma, salivary gland tumors (pleomorphic adenoma, myoepithelioma, polymorphous low-grade adenocarcinoma, epithelial-myoepithelial carcinoma). Hence for a definitive diagnosis, immunohistochemical study was advised.

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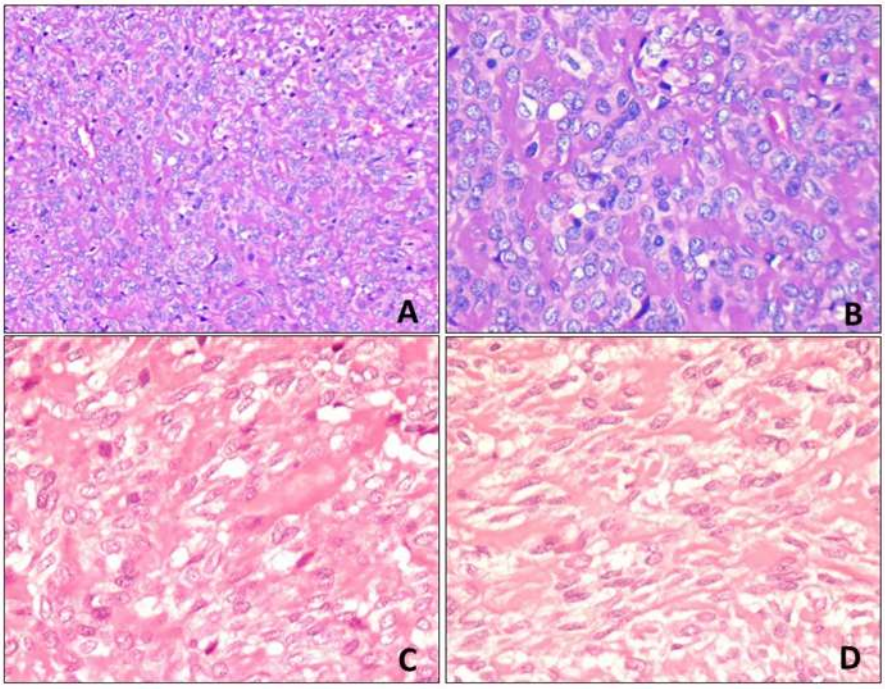
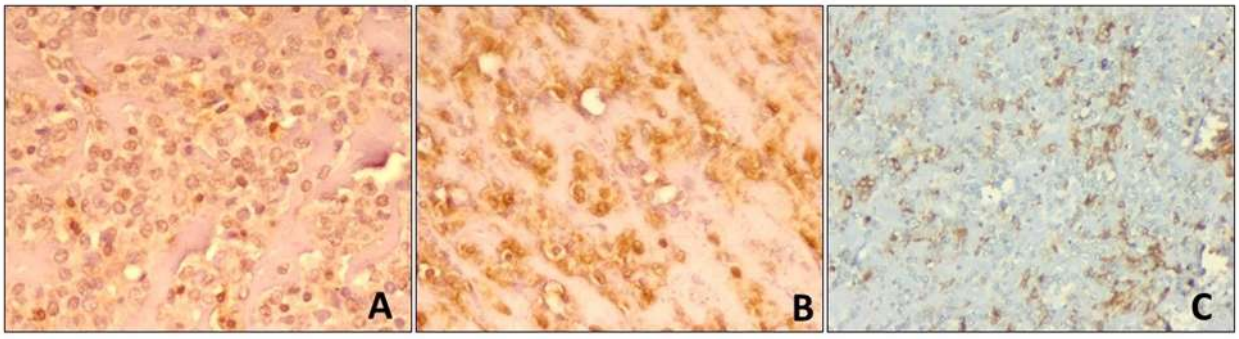
Figure 2: A-B: H&E-stained sections showing solid tumor with round to oval cells with inconspicuous nucleoli and vesicular chromatin, surrounded by scanty, hyalinized stroma [H&E, A-20X, B-40X]; C-D: Sections showing tumor cells with epithelioid morphology [H&E, C&D-40X]



On immunohistochemistry, cells were positive for p63 & S100 and also showed focal positivity for SMA (smooth muscle actin) [Figure 3]. These tumor cells showed negative results with synaptophysin & chromogranin.

Figure 3: A- Tumor cells showing positivity for p63 [40X]; B- Immunoreactivity for S100 [40X]; C- Tumor cells are focally positive for SMA [20X]



Based on an extensive histological evaluation in conjunction with immunochemistry, a definitive diagnosis of Myoepithelioma of Parotid gland was given. The patient recovered from the surgery without complications and her facial nerve was functioning well. The patient had no sign of recurrence at 10 months and is currently being seen regularly for routine monitoring.

Discussion:

Myoepithelioma is a rare benign tumor arising from neoplastic myoepithelial or basket cells. These cells are found between the basement membrane and the basal plasma membrane of acinar cells.[1-3] They are made up of numerous cellular elements including smooth muscle actin, myosin, and intermediate filaments. Myoepithelial cells are supposed to have contractile units that aid in excreting glandular secretions.

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Myoepitheliomas account for only 1% of all salivary neoplasms. Mainly it affects the parotid gland (40%) followed by minor salivary glands (21%) and rarely submandibular gland.[2-4] The complete histological description of myoepithelioma was first explained in 1943.[4-7] Subsequently, myoepitheliomas have been variously named as "parotid clear cell adenoma of possible myoepithelial origin" and "adeno-myoepithelioma." The term "Myoepithelioma of the salivary gland" was first officially recognized as a subtype of salivary neoplasms in 1991.[3-7]

These tumors can be found in nearly all exocrine gland tissues. These sites could be skin, soft tissue, sweat glands, breast, lacrimal glands, Bartholin's glands, nasal septum, nasopharynx, etc.[3-8] The various differential diagnosis to be considered while evaluating these lesions are abscess, mucocele, schwannoma, neurofibroma, leiomyoma, benign fibrous histiocytoma, smooth muscle neoplasms, pleomorphic adenoma, mucoepidermoid carcinoma, myoepithelial carcinoma, etc.[2-8]

We can differentiate these entities based on their clinical & radiological findings. But to arrive at a definitive diagnosis, an extensive histopathological examination is must along with appropriate immunohistochemical markers. Abscess shows presence of dense acute inflammatory cells in a necrotic background. In mucocele, we find abundant mucin, scant to moderate cellularity with cohesive, monolayer clusters and sheets of epithelial cells. No atypia is seen. Benign nerve sheath tumor like neurofibroma shows spindle shaped cells with buckling of nuclei. Schwannoma shows presence of hypercellular and hypocellular areas, termed as Antoni A & B areas. Leiomyoma shows spindle shaped cells in intersecting fascicles with spindled nuclei & vesicular chromatin along with abundant eosinophilic cytoplasm.

Pleomorphic adenoma shows 3 components, comprising of epithelial (ductal) component forming the inner layer of cysts and tubules. Myoepithelial cells as the outer layer of cysts and tubules and scattered within the myxoid stroma. Cytology of myoepithelial cells can be plasmacytoid, spindled, epithelioid, clear or stellate shaped. Stromal component is typically myxoid, chondroid or myxochondroid. It can also be hyalinized or fibrotic. Metaplastic changes may be seen.

Mucoepidermoid carcinoma shows varying proportions of epidermoid cells, intermediate cells and mucocytes arranged in solid nests, sheets or cords of epidermoid cells. Mucous cells are embedded in epidermoid cell nests or lining cystic spaces. Intermediate cells found within epidermoid cell nests or forming separate nests. Background stromal sclerosis is noted. Luminal or extracellular pools of mucin may be present. Mitosis, necrosis and pleomorphism more commonly seen in high grade tumors.

Of all benign major and minor salivary gland neoplasms, myoepitheliomas account for 2.2% and 5.7%, respectively.[6-8] Most of these tumors occur in adults; can be seen in children also. Male and female are equally affected. The average age of a patient with myoepithelioma is 42-45 years, with a range of 9–85 years.[5-8] The most common clinical presentation is a slow growing, painless mass, as seen in the clinical presentation of our patient.

The diagnosis is usually made with a combination of radiologic imaging and tissue histology. CT scan shows a well circumscribed, smooth or lobulated, homogenous enhancing lesions, similar to what was seen in our case.[7-8] Typical MRI studies show a well-defined homogenous isointense and hyperintense mass on T1- and T2-weighted imaging, respectively.

On gross inspection, they have a solid, tan or yellow-tan, glistening cut surface, similar to what was seen in our gross examination. Myoepithelioma shows spindled, epithelioid, plasmacytoid, clear or oncocytic cells. These tumours are well circumscribed or encapsulated with a thin capsule. Stroma, when present, may be hyalinized, fibrous, myxoid or mucoid.

Subtypes of myoepitheliomas are classified by cell morphology: spindle (interlacing fascicles with a stroma-like appearance), plasmacytoid/hyaline (polygonal cells with eccentric nuclei and dense, nongranular or hyaline, abundant eosinophilic cytoplasm), epithelioid (nests or cords of round to polygonal cells, with centrally located nuclei and a variable amount of eosinophilic cytoplasm), and clear (polygonal cells with abundant optically clear cytoplasm, containing large amounts of glycogen. Of those subtypes, spindle cell type is most common (65%) followed by plasmacytoid (20%). Due to histological and cytogenetic similarities, a

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myoepithelioma can sometimes be misdiagnosed as a pleomorphic adenoma. IHC is very helpful in arriving at a final diagnosis. In our case, cells were positive for p63 & S100 and also showed focal positivity for SMA (smooth muscle actin). These tumor cells showed negative results with synaptophysin & chromogranin.

The treatment of choice is surgical excision. However, the role of chemoradiation is not well established due to rare occurrence of this entity. The recurrence rate for myoepitheliomas has been reported to be 15–18%.[6-8] Rarely, malignant transformation can be seen in these tumors. After complete surgical excision, a proper follow-up is needed in the treatment protocol to watch for any recurrence.[5-8] In our patient, no recurrence was found till the last follow-up of one year.

Conclusion:

To conclude, myoepitheliomas commonly involve parotid gland, but its overall occurrence is very less. These are still a rare salivary gland neoplasm. It is difficult to diagnose these tumors on the basis of clinical and radiological findings. Hence, to arrive at a definitive diagnosis, a detailed histopathological evaluation is needed in conjunction with other clinical details. Due to presence of many other differentials at same site, a proper use of immunohistochemical markers is warranted to arrive at a final diagnosis. In our case also, IHC played an important role in labelling the tumor as benign myoepithelioma of right parotid gland.

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