

ORIGINAL RESEARCH PAPER

ENT

A CASE REPORT ON MIXED TUMOR OF SUBMANDIBULAR SALIVARY GLAND AND REVIEW OF LITERATURE

KEY WORDS: Mixed Tumor, Submandibular Salivary Gland

Dr. M. Lakshmi Narayana*

Assistant Professor, Department of ENT, PESIMSR, Kuppam. *Corresponding Author

Dr. N. Sindhuja

Post graduate, Department of ENT, PESIMSR, Kuppam

Dr. Udaya Kumar. M

Professor, Department of Pathology, PESIMSR, Kuppam

ABSTRACT

Pleomorphic adenoma (mixed tumor) is most common in parotid followed by submandibular gland. It exhibits bimodal population of cells with varied stroma. We report a case of 30 years old female presented with left submandibular gland swelling. On examination, a 5x4 cms firm swelling which was ballotable on bidigital palpation was seen. Ultrasound showed well defined hypoechoic vascularised lesion in the left submandibular triangle. FNAC showed features of pleomorphic adenoma. The lesion was excised and post operative period was uneventful. Histopathology showed epithelial and myoepithelial cells distributed in a chondromyxoid stroma without any vascular and capsular invasion. In conclusion mixed tumor of salivary glands should be excised without any delay as the chances of malignant transformation to carcinoma ex pleomorphic adenoma was there.

INTRODUCTION:

Salivary gland neoplasm's constitute around 3% of head and neck tumors. According to literature benign tumors are common in major salivary glands and malignant are common in minor salivary glands. Pleomorphic adenoma is the most common benign salivary gland tumor. The occurrence of pleomorphic adenoma in parotid is common while in submandibular salivary gland is rare. According to Spiro article 8.4% of salivary gland neoplasms constitute submandibular salivary gland neoplasms. Among those 45% were benign and 55% were malignant¹.

Pleomorphic adenoma exhibits a varied histological diversity. It consists of both epithelial and myoepithelial cells in variable background stroma. Stroma in pleomorphic adenoma is mucoid, myxoid, chondroid or hyalinized. This is a case report on submandibular salivary gland pleomorphic adenoma with the review of literature on pleomorphic adenoma of submandibular region.

CASE REPORT:

A 30 years old female presented with left sided upper neck swelling for the past six months. It was painless and progressive. On examination, a 5×4 cm globular swelling was seen in the left submandibular region. The swelling was firm, non tender, not compressible, non-pulsatile and not fixed to the underlying structures with normal overlying skin. Swelling was ballotable on bidigital palpation and transillumination test was negative (fig. 1). USG neck revealed a $5.5 \times 4.0 \times 1.6$ cm well defined hypoechoic lesion in left submandibular gland with a thin rim of normal salivary gland tissue. Vascularity was seen within the lesion without any calcification (fig.2). USG guided FNAC shows features suggestive of pleomorphic adenoma. So, excision of the tumor was done and sent for histopathological examination (fig.3, 4).

Histopathological examination reported a well encapsulated benign neoplasm comprising of bimodal population of cells. Epithelial component comprised of cells arranged in anastomosing tubules, cords and sheets. Cells were round having bland nuclear chromatin and duct lumen contains eosinophilic material. Myoepithelial component comprised of spindle cells with bland chromatin. Chondro-myxoid stroma was noted (fig.5, 6). Also seen were normal salivary gland tissue. No nuclear atypia or increased mitosis was noted. No capsular or vascular invasion was seen. The final diagnosis of Pleomorphic Adenoma of left submandibular gland was made.



FIG1: Pre-operative picture of the patient

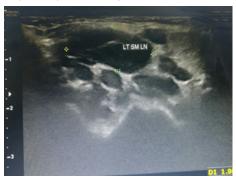


FIG2: USG report showing well defined hypoechoic lesion in left submandibular gland



FIG3: Intra – operative picture showing submandibular gland removal



FIG4: Post – operative picture of the patient with well healed scar



FIG5: Histopathological picture showing chondromyxoid matrix and myoepithelial cells with normal submandibular gland below.

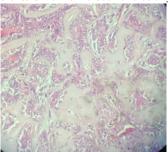


FIG6: high resolution (40X) picture of the submandibular gland showing chondromyxoid matrix and myoepithelial cells.

DISCUSSION:

Pleomorphic adenoma is a mixed tumor possessing both epithelial and myoepithelial elements with varied histological stroma. They are usually seen in middle aged women. These lesions are usually solitary, ovoid, well defined, slow growing and painless masses. The larger tumors may have pedunculated outgrowths from the main lesion due to the presence of pseudopods and seen like multiple masses on examination^{2,3}.

Pathologically pleomorphic adenoma is classified as stroma-rich, cell-rich and classic variants, of which the most common is cell rich variants^{4,5}. Stroma-rich variants were larger than celluar rich variants⁶. The mesenchymal components include myxoid, chondroid, hyaline, fatty and calcified tissue, with most common occurrence of myxoid variant. Our case has a combination of chondroid and myxoid elements as chondromyxoid stroma. The immunohistochemical analysis of pleomorphic adenomas is found to be negative for Ki-67 and P53 which shows a low proliferative rate and have good prognosis⁶. There is no difference in the histological features of pleomorphic adenoma between parotid and submandibular glands.

The differential diagnosis should include basal cell adenoma, adenocarcinoma, polymorphous low grade adenocarcinoma, mucoepidermoid carcinoma and lymphoma. On ultrasound well defined rounded hypoechoic lesions with lobulated or bosselated contour are seen. They may have posterior acoustic enhancement and appear heterogeneous secondary to hemorrhage, calcification and necrosis'. Small tumors are generally smooth and well defined whereas larger tumors tend to be lobulated. CT scan helps in determining the extent of disease and local spread. Intact fat plane distinguishes benign tumors from malignant.

Recurrence of pleomorphic adenoma in parotid was 0.8-5% and in submandibular gland was very rare. Alves et al found 1 case of recurrence out of 54 cases in submandibular pleomorphic adenoma⁶. There are different hypothesis for the recurrence which include pseudopods in the capsule, incomplete capsule formation, satellite nodules, stroma-rich variant, incomplete surgical resection and implantation of tumor during surgery⁸. Pleomorphic adenomas are benign tumors but can turn malignant (carcinoma ex pleomorphic adenoma) in up to 5-25% of untreated cases³. Benign metastasizing pleomorphic adenoma is one more variant which is histologically similar to pleomorphic adenoma but metastases to distant sites is seen. The most common sites of metastases were bone, lung and cervical lymph nodes. The survival was poor in metastasizing pleomorphic adenoma¹⁰. Considering the malignant conversion and metastasizing nature of pleomorphic adenoma, early treatment in the form of surgical excision is recommend.

CONCLUSION:

In our case, an adult female presented with a left submandibular swelling with FNAC suggestive of a pleomorphic adenoma. Surgical excision is advocated without any delay as treatment of choice to avoid malignant transformation of the submandibular gland pleomorphic adenoma in future.

REFERENCES

- Spiro RH. Salivary neoplasms: overview of a 35 year experience with 2,807 patients. Head Neck Surg. 1986;8:177-184.
- Rai S, Sodhi SPS, Sandhu SV. Pleomorphic adenoma of submandibular gland: An uncommon occurrence. Natl J Maxillofac Surg. 2011 Jan-Jun; 2(1): 66–68.
- Subhashraj K. Salivary gland tumors: A single institution experience in India. Br J Oral Maxillofac Surg. 2008;46:635–8.
 Becerril-Ramírez PB, Bravo-Escobar GA, Prado-Calleros HM, Castillo-Ventura BB,
- Becerril-Ramírez PB, Bravo-Escobar GA, Prado-Calleros HM, Castillo-Ventura BB, Pombo-Nava A. Histology of submandibular gland tumours, 10 years' experience. Acta Otorrinolaringol Esp. 2011;62:432–5.
 Mahmood Jahangir Nezhad, Saedeh Atarbashi Moghadam, Sepideh Mokhtari,
- Mahmood Jahangir Nezhad, Saedeh Atarbashi Moghadam, Sepideh Mokhtari, ShirinTaravati. Different Histolopathologic Features of Pleomorphic Adenoma in Salivary Glands. International Journal of Oral & Maxillofacial Pathology; 2013:4(2):07-11.
- Alves FA, Perez DC, Almeida OP, Lopes MA, Kowalski LP. Pleomorphic adenoma of the submandibular gland: Clinicopathological and immunohistochemical features of 60 cases in brazil. Archives of Otolaryngology–Head & Neck Surgery. 2002;128(12):1400-3.
- Dumitriu D, Dudea SM, Botar-Jid C, Băciuţ G. Ultrasonographic and sonoelastographic features of pleomorphic adenomas of the salivary glands. MedUltrason. 2010 Sep;12(3):175-83.
- Inan S, Aydın E, Babakurban ST, Akçay EY. Recurrent Pleomorphic Adenoma of the Submandibular Gland. Turkish Archives of Otorhinolaryngology. 2016;54(1):43-46.
- Bhat VS, Biniyam K, Aziz AA, Yeshwanth SK. Carcinoma ex-pleomorphic adenoma of submandibular salivary gland: A case report and review of literature. J NTR Univ Health Sci 2017-6185-8
- Knight J, Ratnasingham K. Metastasising pleomorphic adenoma: Systematicreview. Int J Surg. 2015 Jul; 19:137-45.