

Case Report

Pleomorphic Adenoma of Submandibular Gland: A Rare Occurrence

Dr. Jatin Verma^{1*}, Dr. V.P. Narve², Dr. Mohit Punjabi³, Dr. Vikram Singh Rathore⁴, Dr. Aprajita Pandey⁵

¹ Resident PG, Department of ENT, G.R Medical College & J.A Group Of Hospitals, Gwalior (M. P.)

² Associate Prof. & Head, Department of ENT, G.R Medical College & J.A Group Of Hospitals, Gwalior (M. P.)

³ Resident PG, Department of ENT, G.R Medical College & J.A Group Of Hospitals, Gwalior (M. P.)

⁴ Resident PG, Department of ENT, G.R Medical College & J.A Group Of Hospitals, Gwalior (M. P.)

⁵ Resident PG, Department of ENT, G.R Medical College & J.A Group Of Hospitals, Gwalior (M. P.)

*Corresponding author

Dr. Jatin Verma

Email: drjatinverma@gmail.com

Abstract: The pleomorphic adenoma of the submandibular gland is rare and can present difficulty in diagnosis. The heterogeneous histology, a possible malignant transformation, an incomplete capsule that can determine a recurrence, are the most important characteristics of pleomorphic adenoma. A complete surgical removal of the tumor is the treatment of choice. This case report presents a histologically proven pleomorphic adenoma involving submandibular gland. The case was treated surgically and followed up for more than six months without any recurrence.

Keywords: Pleomorphic adenoma, salivary gland, submandibular gland, tumour

INTRODUCTION

Salivary gland tumors are rare and make up to 3% of head and neck tumors [1]. Approximately 90% of the benign neoplasms of the major salivary gland are associated with the parotid gland. Pleomorphic adenoma (PA) comprises 80–90% of these benign parotid neoplasms. PA of the submandibular and sublingual gland is quite uncommon and comprises rest (8–10%) of the group [2]. Pleomorphic adenoma (PA) is characterized by great histological diversity and myoepithelial cells are considered responsible for

production of the extracellular matrix. This case report presents a case of a histologically proven PA involving the submandibular gland. The case was treated surgically and followed up for more than 6 months with no recurrence.

CASE REPORT

A 52 year old male presented with swelling below right side of jaw for 5-6 years as shown in Figure(1a&1b).



Fig. 1a&1b: Facial Profile showing swelling in submandibular region

Initially the swelling was small in size which increased progressively. It was not associated with pain, fever, loss of weight, loss of appetite or related to meals. There was no history of exposure or contact with tuberculosis. Physical examination revealed a right submandibular mass measuring 5x3cm with well defined margins. The mass was non-tender, firm on palpation with a nodular feel and normal overlying skin.

The swelling was mobile in horizontal and vertical planes and was not attached to overlying skin and underlying tissues. The swelling was non-compressible and non-fluctuant. The cervical lymph nodes were not palpable in the neck. The floor of the mouth and Wharton's duct showed no abnormality. Other systemic examinations were unremarkable. Fine needle aspiration revealed cellular smears composed of a

mixture of epithelial and stromal components. The epithelial cells were arranged in loose sheets as well as singly distributed displaying plasmacytoid nuclei with bland nuclear chromatin and dense cytoplasm. The stromal component was composed of fibrillary chondromyxoid substance admixed with spindle-shaped mesenchymal cells. These cytological features are consistent with Pleomorphic Adenoma. USG NECK showed a hypoechoic lesion of size 4.6x2.8 cm with cystic changes in it arising from submandibular gland with well defined lobular margin. Axial and coronal views of the Computed Tomography (CT) scan showed a moderate sized with lobulated surface, mild heterogenous enhanced soft tissue density lesion in right submandibular region without perilesional soft tissue stranding and bone erosion and lymphadenopathy suggesting most likely possibility of Benign mixed tumor of right submandibular gland (Figure 2).



Fig. 2: Axial CT Scan

A provisional diagnosis of Pleomorphic Adenoma was made and the patient was posted for submandibulectomy. With a standard submandibular incision, the submandibular gland and the mass was excised and sent for histopathological examination. Primary closure was done in a layer-wise manner with a drain attached (Figure 3 & 4).



Fig. 3: Intraoperative Photo



Fig. 4: Excised lobular tumor

The histopathological examination showed darkly stained tumor cells lying in a predominantly mesenchyme like background and these findings were confirmatory of Pleomorphic Adenoma (Figure 5).



Fig. 5: Histopathology Slide

DISCUSSION

Pleomorphic Adenoma is an epithelial tumor of complex morphology, possessing epithelial and myoepithelial elements intermingled with mucoid, myxoid, or chondroid tissue arranged in a variety of patterns and embedded in a mucopolysaccharide stroma [3]. It is the commonest benign tumor of salivary glands [4] and accounts for 90% of all salivary gland tumors. The submandibular gland is the second most common site of Pleomorphic Adenoma after the parotid gland [5]. It is also the most frequent benign tumor arising in submandibular gland [6]. The differential diagnosis should include basal cell adenoma, adenocarcinoma, mucoepidermoid carcinoma and lymphoma.

CT scan or magnetic resonance imaging (MRI) are the gold standard radiological tools for lesion arising from the major or minor salivary glands. Adjunctive procedures like ultrasound guided needle aspiration or fine needle aspiration are non-confirmatory. An incisional biopsy can be taken initially if the lesion is of large size. The recommended surgical approach is with a direct submandibular incision which provides an easy access. The excision of the tumor should also be

accompanied by the removal of the submandibular gland in toto. Incomplete removal of the glandular tissue paves the way for a definitive recurrence. Pleomorphic Adenomas are benign tumors with a well-documented transformation to malignancy (carcinoma ex pleomorphic adenoma). It is estimated that up to 25% of untreated PAs undergo malignant transformation [7]. Therefore, early definitive treatment is strongly recommended.

CONCLUSION

A high index of suspicion is required for non-inflammatory lesions in the submandibular region to avoid undue delay in definitive treatment. Preoperative diagnosis with FNAC has proven to be highly accurate. We advocate surgical excision because of chances of malignant transformation later in life.

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