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Case Report

Pleomorphic adenoma of the submandibular gland: a case report

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Abstract

Salivary gland tumors are relatively rare and constitute only about 1-4 % of head and neck tumors. Pleomorphic adenoma (PA) is the most common benign tumor of salivary glands. Approximately 80% of pleomorphic adenomas occur in the parotid gland, rest 10-20% in submandibular and minor salivary glands. Here, we present a confirmed case of pleomorphic adenoma of the submandibular gland.

Introduction

Salivary gland tumors are found less in the population and comprise only 1-4% of head-face-neck tumors, most of which are benign. Pleomorphic adenoma comprises majority (about 80%) of the benign tumors of salivary glands [1]. It can also be classified as a mixed tumor since it has both epithelial and mesenchymal components as seen on histology [2]. Majority of pleomorphic adenomas occur in the largest salivary gland, i.e., the parotid (85%), 5% in the submandibular gland, and 10% in sublingual glands and minor salivary glands [3]. Here, we describe a histologically proven case of submandibular gland pleomorphic adenoma and its management, with no recurrence on regular follow up.

Case presentation

A 32-year-old male presented to Otorhinolaryngology OPD, ESIC medical college hospital with a history of firm palpable mass in the right submandibular region [Fig-1a and 1b], for the past 4 years.

According to the patient's information, the swelling was insidious in onset, slow growing in nature and painless, but for the last 1 year it had progressed rapidly. There was nothing significant in past medical and personal history. Also, there was no history of difficulty in swallowing, breathing, or change of voice.

On inspection, the swelling was located in the right submandibular region, oval in shape and measured approximately 7 cm × 6 cm. When palpated, the mass was firm in consistency, non-tender, mobile with well-defined borders and normal overlying skin, not fixed to the underlying tissue. Swelling was ballotable on bimanual palpation. Patient did not have any neurogenic or functional deformity. No reduction in salivation from right Wharton's duct was seen on intraoral examination. No significant lymphadenopathy was present. Examination findings of oral cavity and oropharynx were within normal limits. Nothing significant was found on indirect laryngoscopic examination.

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On the basis of history and examination, tumour of right submandibular gland was suspected. Computerized tomography (CT) scan of neck was done to determine the extent of the lesion. Axial and coronal cut sections of CT scan showed evidence of a large lobulated, heterogeneously enhancing mass lesion involving the right submandibular gland (approx. 6.5cm× 5.4cm) [Figure-2a and 2b]. Fine needle aspiration cytology (FNAC) was done. The smear showed high cellularity, arranged in cohesive clusters and sheets. Myoepithelial cells including plasmacytoid cells, basaloid cells, along with stromal fragments were seen, suggesting pleomorphic adenoma.

Based on the above mentioned findings, en bloc resection of right submandibular gland, along with the tumour [Figure-3a], was performed under general anaesthesia, with precise right submandibular incision. Histopathological examination report of the excised tumour showed, tubules and cords lined by inner epithelial cells and outer myoepithelial cells, against chondro-myxoid stroma [Figure-3b]. No evidence of malignancy or dysplasia was noted. Therefore, the diagnosis of pleomorphic adenoma of right submandibular gland was confirmed. Post operative period was uneventful, without any complication. There were no signs of recurrence on regular follow up.

Figure-1: *Photograph showing right submandibular mass. Fig-1a: Frontal view and Fig-1b: Lateral view*

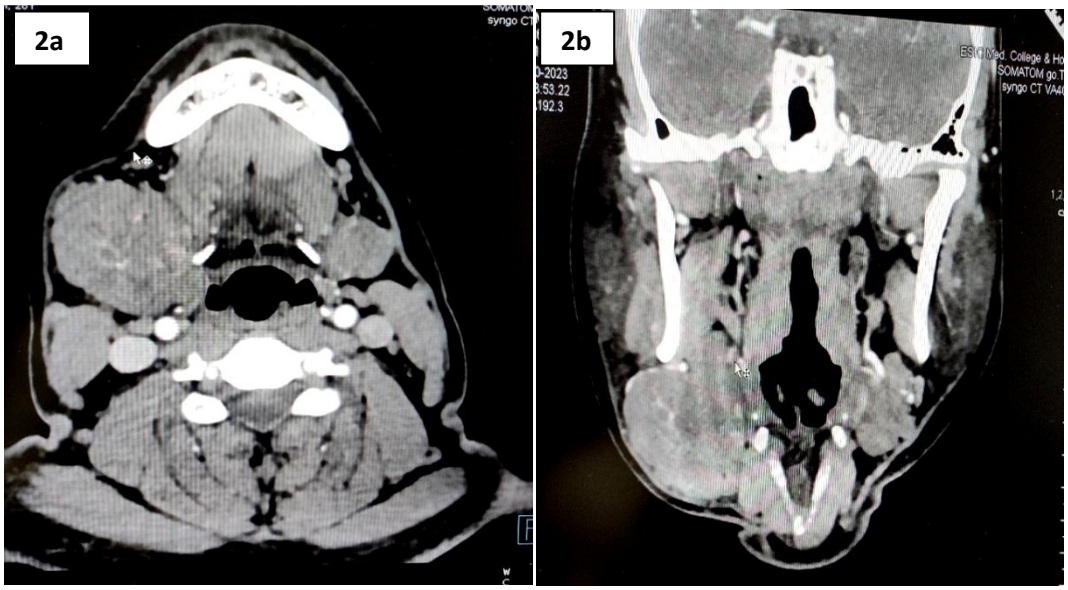

Figure-2: *Axial (2a) and coronal (2b) cut sections of CT scan showing well defined heterogeneously enhancing mass of right submandibular gland.*

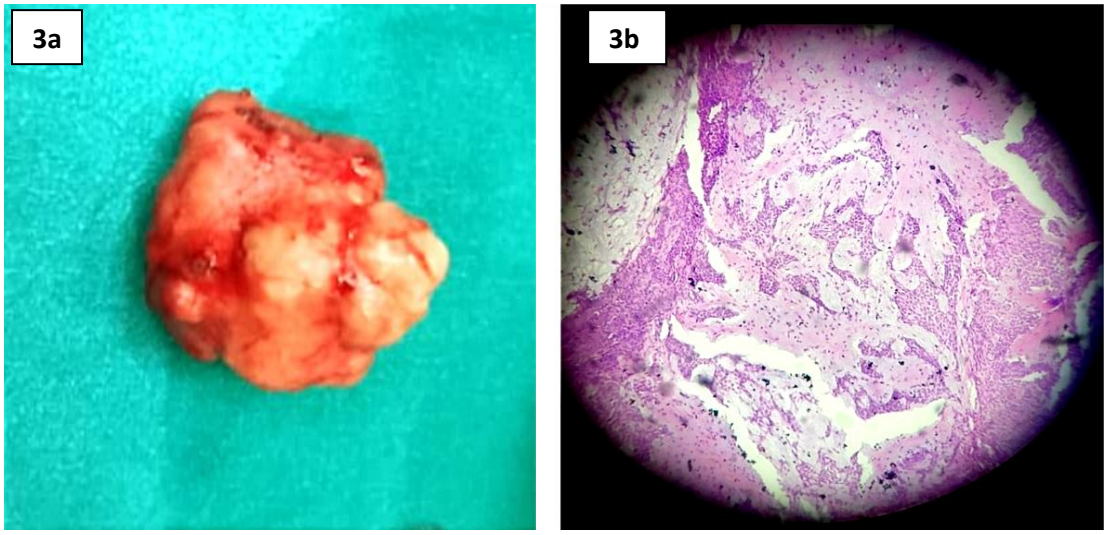

Figure-3: *Photograph of gross specimen of resected adenoma (Fig-3a).* Histopathological examination of the mass *showing epithelial and myoepithelial cells against chondro-myxoid stroma (Fig-3b)*

Discussion

Submandibular glands are next to parotid, in reference to site of occurrence of PA [4]. The common tumours of the submandibular glands, in descending order are: PA (36%), adenoid cysticcarcinoma (25%), mucoepidermoid carcinoma (12%) and malignant mixed tumor (10%) [5]. The differential diagnosis should include basal cell adenoma, adenocarcinoma, mucoepidermoid carcinoma and lymphoma. Pleomorphic adenomas are benign mixed tumours as they have complex histological features.

Pleomorphic adenoma is an epithelial tumor of complex morphology, possessing epithelial and myoepithelial elements mixed with mucoid, myxoid, or chondroid tissue arranged in a variety of patterns and embedded in a mucopolysaccharide stroma [6].

In a 10 year study conducted by Becerril-Ramírez *et al.*, 22 cases of submandibular gland neoplasms were noted, in which 19 cases (86%) were benign and 3 cases (14%) were malignant. The mean age of occurrence of PA was 39.8 years with female to male ratio of 3.5:1 [7]. Rapidis *et al*. Studied clinicopathologic featuresof 23 patients with submandibular gland tumors, out of which 9 were benign and 14 were malignant. They found that PA is most common benign tumor and present a mild course of disease [8].

PA mainly presents as a painless, slow-growing, firm, mobile mass in the submandibular triangle without fixation to the floor of the mouth or the mandible. There is no lymphadenopathy and nerve involvement are rarely seen. Ultrasonography helps in differenting solid and cystic lesions of the salivary glands [9]. Fine needle aspiration cytology (FNAC) and core biopsies are helpful in diagnosis. Radiological investigations such as CT and MRI are gold standard tools for differentiating lesions arising from the major or minor salivary glands.

FNAC is the most essential and important modality for the pre-operative diagosis of salivary gland tumours. The accuracy for the diagnosis of benign or malignant salivary gland tumour by FNAC is around 80–90% [10]. It also helps to differentiate between tumours and inflammatory conditions or enlarged lymph nodes. In a study of 25 submandibular neoplasms by Ethunandan et al., FNAC accurately identified 78% of benign tumors but no malignant tumors [11]. Song IH et al., compared the superiority of core biopsy over FNAC for diagnosing salivary gland tumours in terms of adequacy (97.4% vs. 93.8%), sensitivity (88.2% vs. 58.2%), specificity (99.4% vs. 98.6%), PPV (97.8%

vs. 88.9%), NPV (96.6% vs. 92.6%), and accurate tumour subtyping (88.3% vs. 70.7%) [12].

Pleomorphic adenomas are well demarcated from the surrounding tissue by a pseudocapsule due to compression of the surrounding parenchyma and fibrosis. This fibrous capsule has to be completely excised to prevent recurrence [13].The tumour should always be completely excised alongwith the submandibular gland.The incidence of malignant transformation in adenomas (carcinoma ex pleomorphic adenoma) ranges from 1.9 to 23.3% [14]. The risk increases with a long history of appearance, recurrence, the advanced age of the patient, and location in the major salivary gland [15].

Conclusion

Pleomorphic adenoma of the submandibular gland is a rare entity and should be diagnosed meticulously. Complete surgical excision with removal of the fibrous capsule, is the current treatment modality. Proper diagnosis and surgical management at appropriate time, gives good results regarding cosmetics and recurrence of the disease.

Informed Consent

Informed consent was obtained from the patient for publishing his data, for furthur research purpose.

Conflict of Interests: None

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