CASE REPORT

PLEOMORPHIC ADENOMA OF SUBMANDIBULAR SALIVARY GLAND - A CASE REPORT

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How to cite this article

Dinesh Y, K. Ramalingam, P. Ramani, Vedha V, Mahathi N. Pleomorphic Adenoma of Submandibular Salivary Gland - A Case Report. Int J Histopathol Interpret 2022; 11(1):23 – 28.

Received: 20-05-22; Accepted: 21-06-22; Web Published: 30-06-2022

Abstract:

Pleomorphic adenoma is a benign salivary gland neoplasm with its most common site of origin being the parotid gland, usually involving the superficial lobe of the parotid gland. Pleomorphic adenoma is a painless swelling which gradually increases in size if left untreated. It has a female predilection and occurs within the age range of 30-60 years. Here we report a rare case of a pleomorphic adenoma of the right submandibular gland presented by a 33 years old how it was diagnosed using cytology, histopathology and managed by surgical excision have been described.

Keywords: Pleomorphic adenoma, Salivary gland tumor, Submandibular gland, Surgical excision.

INTRODUCTION

Pleomorphic Adenomas (PA) or benign mixed salivary gland tumors are the most common neoplasms that arise in both major and minor salivary glands. According to Pinkston 1999, approximately 75% to 85% of all PAs occur in the parotid gland, with only 8% arising in the submandibular gland. Those occupying the minor salivary glands represent 7-15% of all reported cases (1). This entity affects any age group but is most commonly seen in the 4th to 7th decade of life and has a female predilection (2). Among the salivary gland tumors occurring in children, pleomorphic adenoma is the most common (3). PAs occur among all age groups, with the incidence rate of about 3.5/100,000. Among intraoral salivary glands, PA affects the palate most commonly (42.63%), followed by the lip (10%), buccal mucosa (5.5%), retromolar area (0.7%), and the floor of the mouth(4). While a majority of such tumors are found in parotid glands (80%), the percentage of those involving submandibular glands is reportedly only (12%). Therefore submandibular PA is considered rare (5) (6).

CASE REPORT

A 33 years old male patient reported to the department of oral medicine and radiology at Saveetha Dental College and Hospital in Chennai, Tamil Nadu, India. The patient came with a chief complaint of swelling in his right neck region for 1 1/2 years. Patient gives a history of swelling which was asymptomatic and initially small in size and increased gradually with time to reach with which he reported to us. He had no relevant medical, facial or past dental history. On extraoral examination, there was a marked swelling in the right neck region. An extra-oral, well defined solitary swelling was evident extending from the superior angle of the mandible, inferiorly to the thyrohyoid region. Facial and neck muscular movements were normal (Fig 1). On palpation, the swelling was firm, non fluctuate, non tender and movable. On intraoral examination, dental caries was evident in relation to 46 and was advised for extraction. Based on the above clinical examination, provisional diagnosis of salivary gland neoplasm- Pleomorphic adenoma was suggested. FNAC was done which showed a 1 ml of serosanguineous aspirate which on H&E staining revealed a numerous round to ovoid cells with eccentrically placed nuclei and eosinophilic cytoplasm suggestive of plasmacytoid cells, few clusters of squamous and few cells resembling mucous cells with clear to foamy cytoplasm admired within a fibromyxoid background. Few squamous and inflammatory cells infiltrate predominantly neutrophils are evident suggestive of salivary gland neoplasm (Fig2).

The results of the patient's routine blood investigations were within normal limits. Intraoral hard tissue examination revealed no anomalies of the teeth in relation to the lesion. The orthopantomogram did not reveal pathological changes in the bone structures. On CBCT examination a hyperdense area was evident on the submandibular gland region, suggestive of salivary gland pathology (Fig3).

Due to the clinical examination, outlook, and history of the lesion, we decided to surgically excise the lesion under general anesthesia and the entire tumour was excised along with the submandibular gland. An informed consent was obtained and the patient was taken under general anaesthesia. Under all aseptic precautions, the usual painting and draping was carried out and the incision line was marked.

A marginal mandibular incision was made using a No. 15 Surgical Bard-Parker® blade. An incision was taken in the preauricular region and extending inferiorly through the skin and the platysma. Soft tissues were bluntly dissected in a retrograde manner in order to prevent injury to the underlying vital structures. The flap was reflected, and the whole encapsulated tumor mass was excised with the boundary line localized in the surrounded healthy tissue (Fig4). Hemostasis was achieved and wound closure done using 3-0 silk.

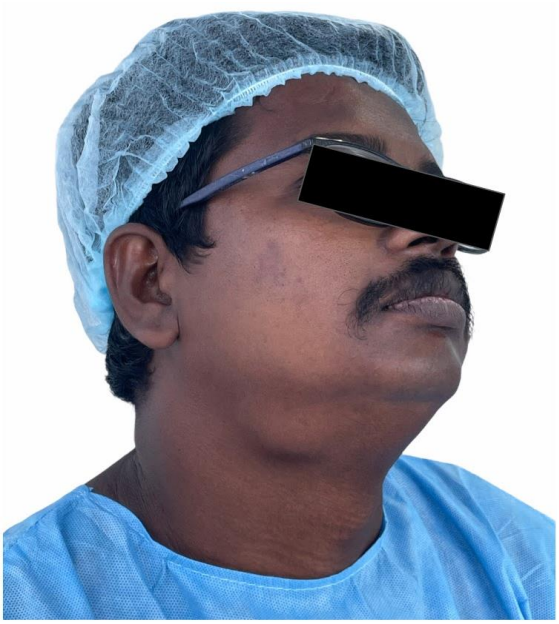

Figure 1: Extraoral image of patient with swelling in the right neck region.

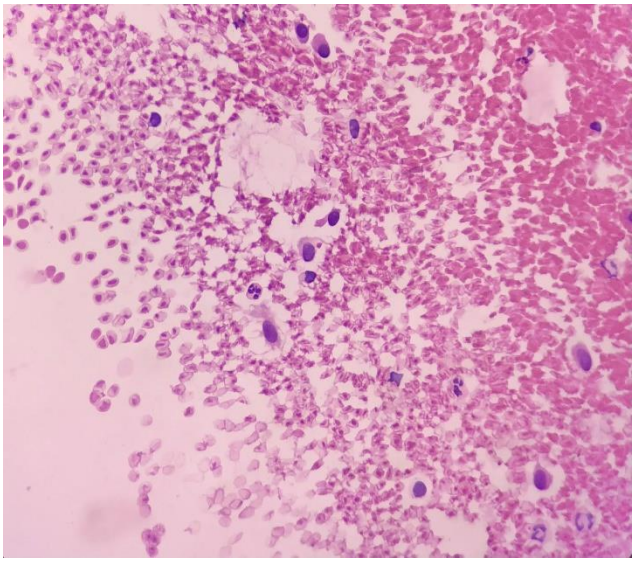

Figure 2: Photomicrograph showing the aspiration cytology (H&E stain, 10x)

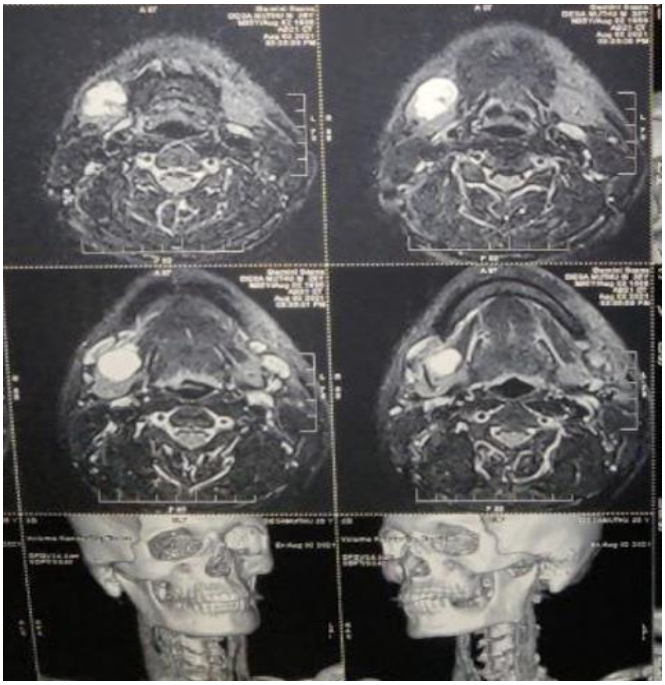

Figure 3: CT pictures showing the lesion

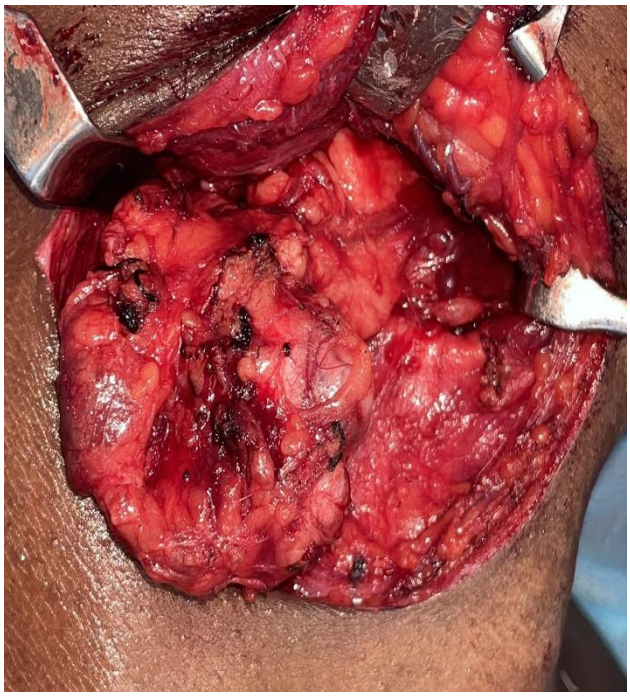

Figure 4: Intra-operative image showing surgical resection of submandibular salivary gland

The excised mass was sent for histopathological evaluation. (Fig 5) This excised tumor mass measured approximately 5.3x2.6x2.3 cm with irregular surface. Histopathological examination revealed the presence of a tumor surrounded by a fibrous capsule. Macroscopically the tumour was indurated, grey black in colour and firm in consistency (Fig6). The entire bit was cut into multiple bits and all the bits were kept for processing.

On histopathological examination, multiple sections showing salivary gland tissue with capsule composed predominantly of myxoid component and epithelial component. Several areas of delicate basophilic stroma with angular cells suggestive of myxoid stroma along with numerous duct like structures with centre lumen filled with eosinophilic coagulum, lined by cuboidal to columnar cells, merging into the myxoid stroma. Several areas showing small to large cystic spaces containing eosinophilic material in the connective tissue stroma are evident.Few angular/ spindle cells with rounded eccentrically placed nucleus and deeply eosinophilia cytoplasm resembling plasmacytoid cells are seen. Several areas showing hyalinization and few basophilic areas with degeneration resembling cartilage?. Correlating clinically histopathology is suggestive of Benign Salivary Gland Neoplasm - Pleomorphic Adenoma (Fig7). Few areas showing normal salivary gland acinar and ductal structures. Adipose tissue, rich vascularity and areas of hemorrhage are evident. Patient is kept under follow up.

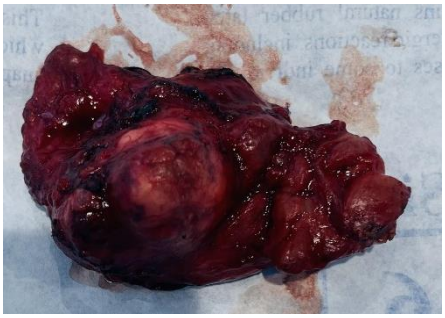

Figure 5: Macroscopic image of resected submandibular salivary gland

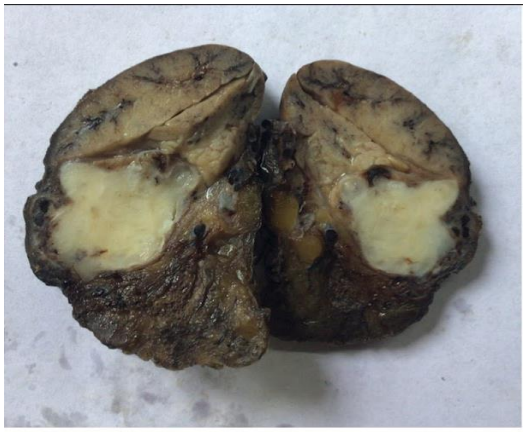

Figure 6: Gross examination of the excised specimen

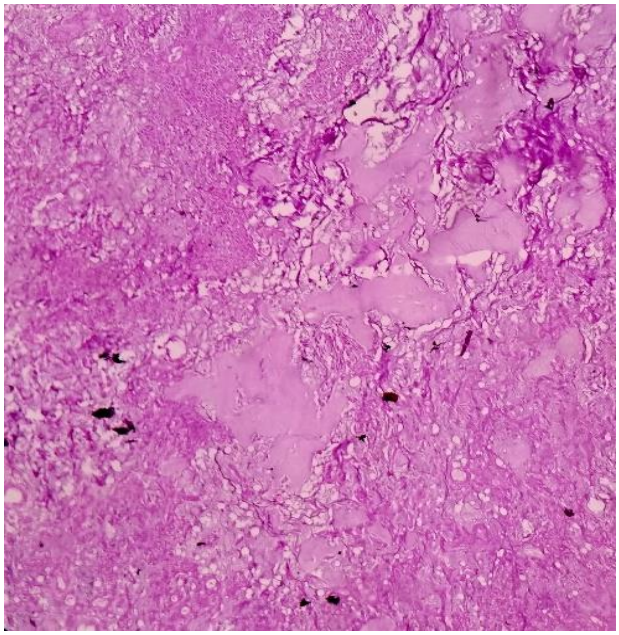

Figure 7 : Photomicrograph showing features of pleomorphic adenoma (H & E stain, 10x)

DISCUSSION

It is rare to report a pleomorphic adenoma- benign salivary gland tumor in the submandibular gland, only a few cases have been reported (7). Being painless and slowly growing, some patients may neglect it to such impressively large sizes especially if surgery was a psychological setback for them. These tumors are usually painless, smooth, firm and asymptomatic until they attain major sizes mostly in the form of pressure symptoms. Imaging studies such as CT and/or MRI can give a reasonable diagnosis. It is usually well demarcated from the surrounding tissue by a pseudocapsule due to compression of the surrounding parenchyma and fibrosis. If this fibrous capsule is completely excised, these tumors could be cured with surgery. In huge sizes they could develop a variegated appearance with areas of hemorrhage, necrosis and calcification. Giant tumors commonly have a lobulated delineation, which supports the diagnosis.(8-10)

At the histological level, PAs demonstrate several phenotypes. Epithelial cells are arranged in sheets and islands showing typical ductal structures lined by epithelial cells with surrounding myoepithelial cells with heterogeneous features such as spindle, squamous, clear, basaloid, oncocytic and sebaceous. The stroma characteristically is diverse with fibrous, chondroid, myxoid or hyaline aspects (8) .The incidence of malignant transformation in PA ranges from 1.9% to 23.3% (5). Risk of the recurrence in salivary gland tumors increases with increasing age of the patient. Our patient did not have any malignant features, as the tumor included only benign epithelial and myoepithelial tissues intermixed with myxoid components.

The treatment of PA is essentially surgical excision (9). Because these tumors are radioresistant, radiation therapy is contraindicated (10). Though these benign tumors are well encapsulated, resection of the tumor with an adequate margin of grossly normal surrounding tissue is necessary to prevent local recurrence because these tumors are known to have microscopic pseudopod-like extensions into the surrounding tissue due to "dehiscence" in the capsule (11).

The recurrence of PA is attributed to implantation from capsule rupture, islands of tumor tissue left behind after surgery, and the multicentric nature of PA. Therefore, long-term follow-up is required (12). Recurrence of tumors is possible especially if the surgical margins are involved, that's why it should be managed through complete excision of the tumor with tumor free surgical margins.

Our case report highlights the problem of many patients especially in the developing countries, who neglect such conditions till it reaches a significant size. Patient education should be enhanced so that they report promptly for necessary interventions.

CONCLUSION

Pleomorphic Adenoma of the submandibular gland is a rare entity, usually seen in adult patients. The most common symptom is a slow-growing, painless submucosal mass on the submandibular gland region. Definitive diagnosis lies in the cytopathological and histopathological examination, and treatment is by surgical excision with wide margins.

Financial support and sponsorship

Nil

Conflicts of interest

There are no conflicts of interest

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