Case report

Solitary Pleomorphic adenoma of the palate

ABSTRACT

Pleomorphic adenoma (PA) is the most common tumor of salivary glands .The parotid gland is the most common site (90%) of origin and it is rarely seen in other minor salivary glands. Nearly 8% of PA involves minor salivary glands- the palate (60-75%) followed by lips and other sites. We report a palatal mass in a 52-year-old gentleman. The patient's major concern was swelling in his oropharynx with voice change and dysphagia. History revealed, the swelling was painless and gradually grew over two years to its present size. The initial diagnosis of pleomorphic adenoma was confirmed upon histopathology reports after incisional biopsy. The case was treated successfully by creating a mucoperiosteal flap under local anaesthesia after tracheostomy. No recurrence till date have been reported after follow up of several months. Pleomorphic adenoma of the palate is a rare tumor, and thus a high index of suspicion and a long-term follow-up is required.

Keywords: pleomorphic adenoma, minor salivary gland tumor, palate

INTRODUCTION

Pleomorphic adenoma (PA) is a mixed tumor composed of epithelial and myoepithelial cells arranged in various patterns, distinguished from the neighbouring tissues by a fibrous capsule. PA is a salivary gland tumor that affects both the major and minor salivary glands and accounts for 40% to 70% of all tumors[1]. The parotid gland is the mostly affected amongst major salivary glands. PA has a female predilection (60%) usually seen in the fourth, fifth, and sixth decades of life[2]. Among intraoral salivary glands, PA affects the palate(42.63%), lip (10%),followed by the buccal mucosa (5.5%), retromolar area (0.7%), and floor of the mouth [3].The uniqueness ascribed in this case is due to its delayed presentation with large lobulated mass intraorally with dysphagia and change in the voice.

CASE PRESENTATION

A 52-year-old gentleman patient reported to the department of Department of Otolaryngology of our hospital . The patient's major complaints were intraoral swelling. Furthermore, history revealed that the swelling was painless and gradually grew over two years to its present size. Additionally, other symptoms including (e.g.dysphagia, change in voice or breathing difficulties) due to the lesions were noted. There was no history suggestive of trauma, fever, or similar swelling elsewhere in the body. Past medical history revealed the patient was healthy and had no co morbidities.

On general physical examination, the patient was averagely built and well nourished. The extra oral examination showed no facial asymmetry or cervical lymphadenopathy. The scalp and spine examination were essentially normal.

Intraoral examination

We noted a solitary, ovoid-shaped swelling measuring 4 cm x 5 cm at the junction of right hard and soft palate was crossing the midline pushing the uvula posteriorly and to the left .Originating from the junction of hard and soft palate(Figure 1). It was occupying the oro-pharynx leading to dysphagia and breathing difficulties. The overlying mucosa appeared healthy and mobile. On palpation, the swelling was multilocular, nontender, nonpulsatile, firm, and movable with well-defined margins with intact gag reflex.

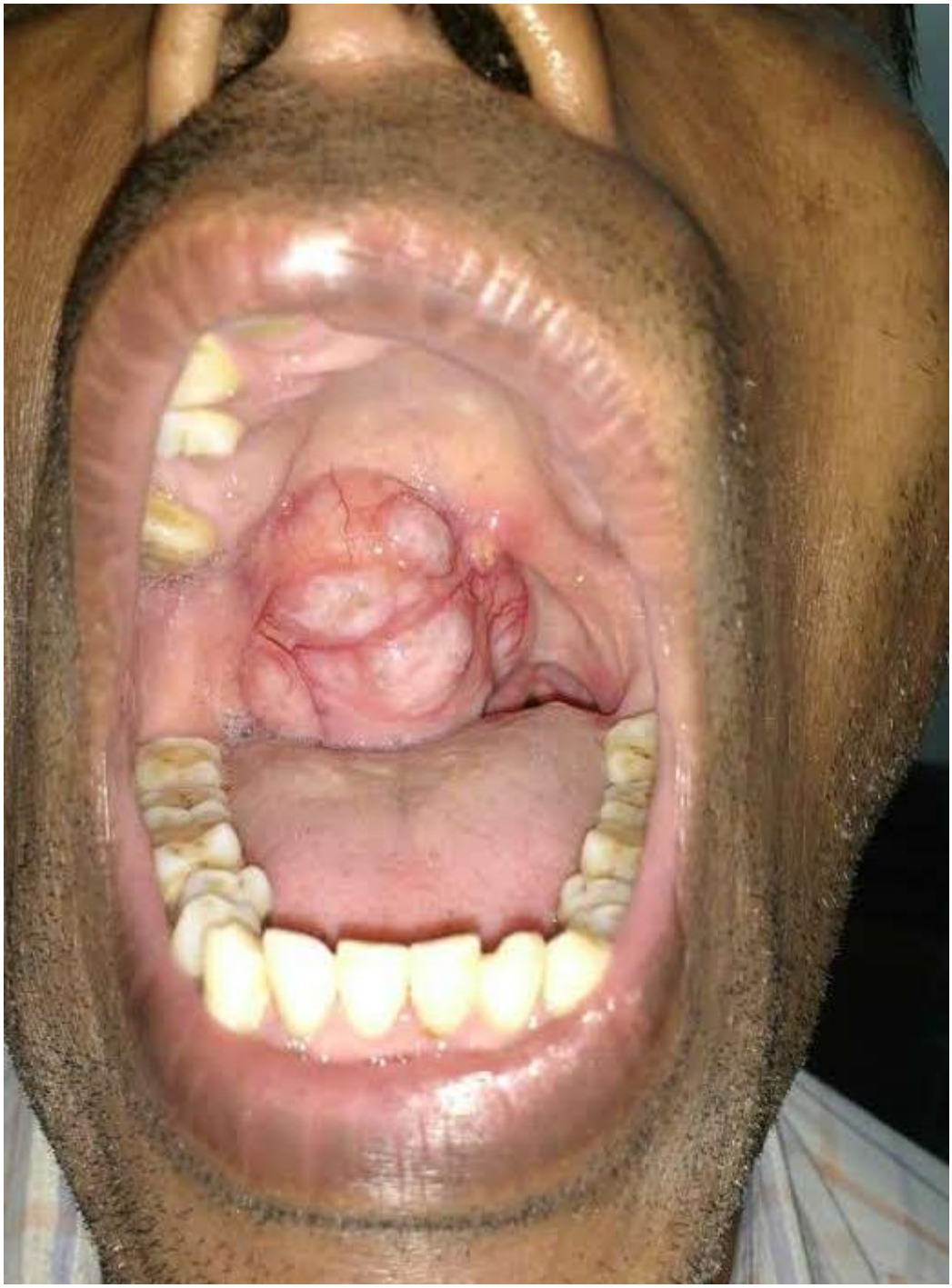

Figure 1: Pleomorphic adenoma, clinical view

Pre-Operative Workup

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. Preoperative punch biopsy was taken and histopathological report suggested pleomorpic adenoma of minor salivary gland. Due to the findings of clinical examination and history of the lesion, we planned to surgically excise the lesion under local anaesthesia. A tracheostomy

under local anaesthesia was performed prior to intraoral excision in-view to protect the airway and early and easy ventilation if this goes wrong.

Surgical Procedure

A horizontal incision was made. The mucoperiosteal flap was raised and the whole encapsulated tumor mass was excised along with the mucoperiosteum using bipolar cautery(Figure 2).The underlying bone was normal with no evidence of erosion of the palate. Haemostasis was achieved and wound closure done using 3-0 vicryl .

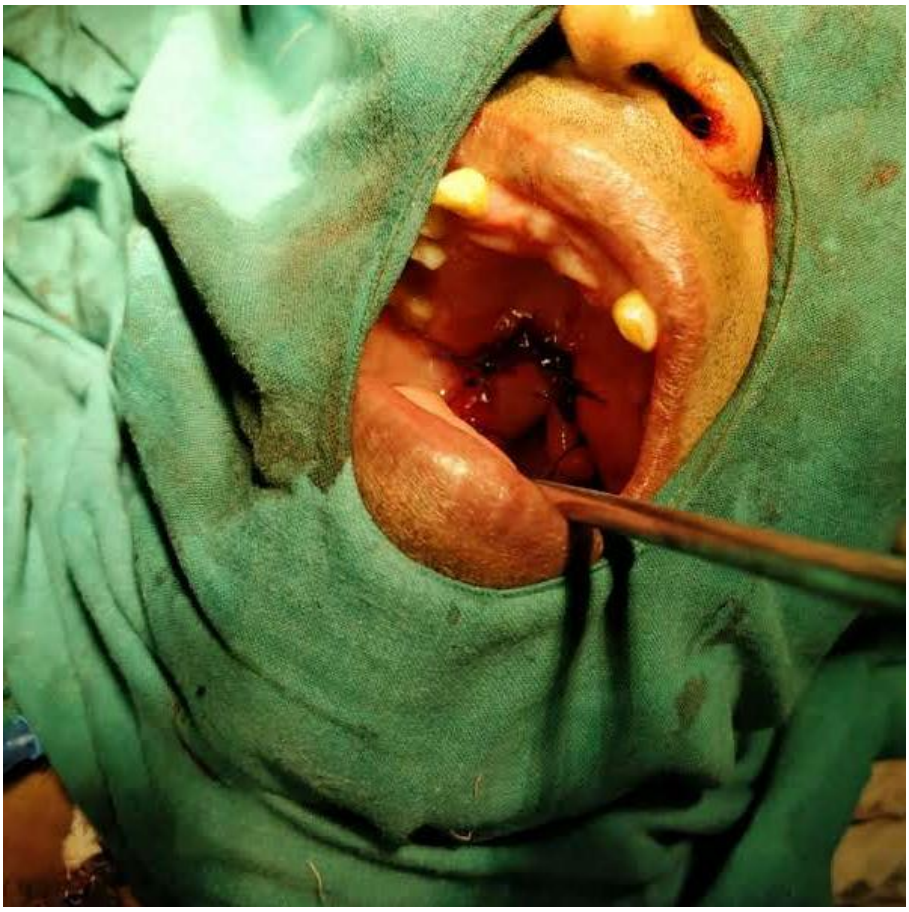

Figure 2: Post resection, intra-operative view

Pathological Findings

The histopathological examination report was suggestive of parakeratinized stratified squamous epithelium along with connective tissue. The underlying tissue was encapsulated with sheets and islands of myoepithelial cells, filled with eosinophilic material.This confirmed the diagnosis of PA.

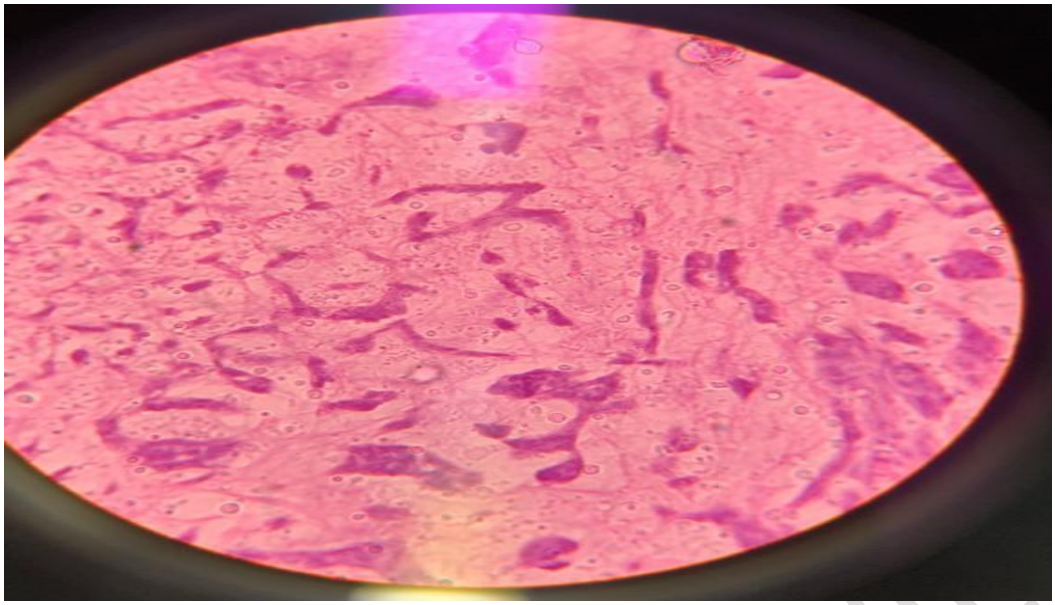

Figure 3: HPE: Myxoid areas representing the mesenchymal component, the epithelial component is seen merging into the mesenchymal element

Immediate Post-Operative and Follow-up

The patient was discharged on the next day with tracheostomy insitu after educating him about the tracheostomy care .The tracheostomy was closed on seventh postoperative day .The patient tolerated the procedure well and recovered uneventfully(Figure 3). The patient was under regular follow-up, and there is no recurrence even after one year of follow-up.

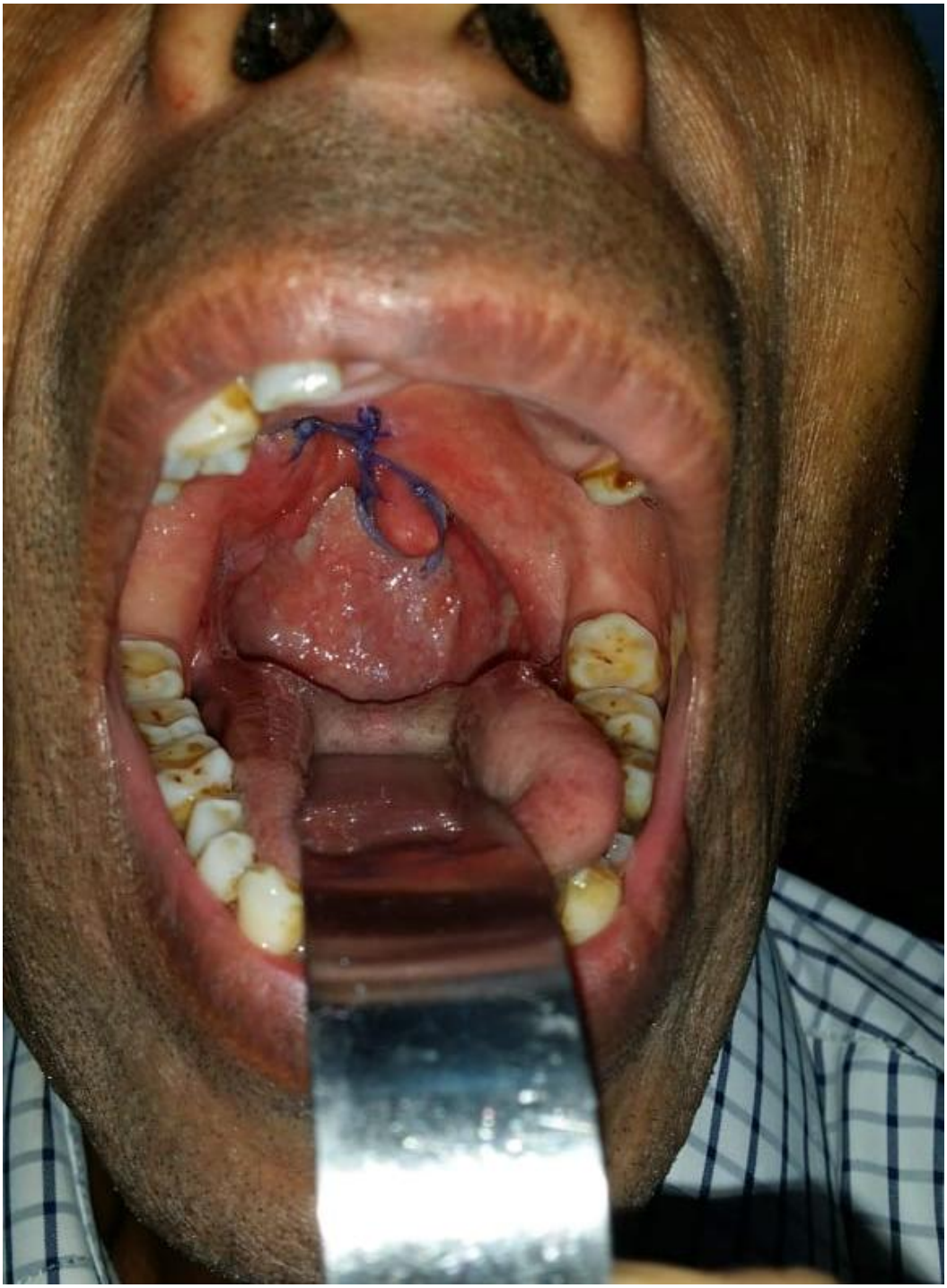

Figure 4 : Recovery,Post-operative day-7

DISCUSSION

PA an intra-oral painless progressive tumor presenting as firm ,dome-shaped swelling on the posterior lateral aspect of the palate [4]. The tightly bound nature of the hard palate mucosa leads to its fixity to underlying structures,whilst tumor of lips and buccal mucosa are freely movable. PA of the palate rarely attains a size more than 1 cm- 2 cm in diameter, as it causes difficulty in mastication, speech, and swallowing[5]. If the overlying mucosa is ulcerated at

presentation, malignancy should be ruled out [1].While the patient in this case presented with large mass of 4-5 centimeters hanging from the roof of the oropharaynx .

Histologically, the tumor contains of myoepithelial, epithelial and mesenchymal components in various proportions with squamous metaplasia and keratin pearls. The epithelial cells can be arranged in sheets or nests of cells giving rise to ductal structures ,glandular appearences with eosinophilic material. Plasmacytoid myoepithelial cells are characterstically seen in minor salivary gland tumors [6] .Additionally ,clear cells, spindle cells and oxyphilic cells may be present. The parenchyma of salivary gland undergoes fibrosis, giving rise to a false capsule [4].

Vuppalapati suggested, PA means architectural pleomorphism visible under light microscopy. PA is also known as mixed salivary gland tumor. The “mixed tumor” aspect accounts for 73% of all salivary gland tumors. The palate is the most common site for PA amongst smaller salivary glands[3].A study of 2078 patients with salivary gland tumor was conducted by Spiro et al ,reported that 20% -40% of tumors arise from minor salivary glands

Daryani et al. reported a case of PA of palate making differential diagnosis of: mucocele, hematoma (bluish discoloration), necrotizing sialometaplasia, adenoid cystic carcinoma ,mucoepidermoid carcinoma,and polymorphous low-grade adenocarcinoma[4]. Sharma et al. reported a swelling with the differential diagnosis of neuroma, neurofibroma and palatal abscess [1].

The treatment of PA is essentially surgical[7-8]. As these tumors are radioresistant, radiation therapy is not indicated [9]. With genetic association of PLAG1, SOX10 and Myb protein expression in various salivary gland neoplasm tissues[10]. As these benign tumors are well-encapsulated, excision of the tumor with safety margins of grossly normal neighbouring tissues is essential to prevent local recurrence. PA are known to have microscopic pseudopod-like extensions into the surrounding tissue due to "dehiscence" in the capsule leading to its recurring nature. The recurrence of PA is due to implantation from the capsule rupture,its multicentric nature and islands of tumor tissue left behind after resection. Therefore, long-term follow-up is necessary.

CONCLUSION

PA of the palate is seldom seen in adults. It presents as a slow-growing, painless ovoid submucosal mass on the hard palate but it rarely exceeds 2 cm in size . Definitive diagnosis is usually after histopathological examination or excision biopsy which may show Myxoid areas representing the mesenchymal component, the epithelial component is seen merging into the mesenchymal element or epithelial component showing small nests.

Consent

Consent was taken from the patient for publication of the case and the photos.

Conflict of Interests

None

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