

Pleomorphic adenoma involving minor salivary glands of upper lip: A rare entity

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Abstract

Benign mixed tumor or Pleomorphic adenoma (PA) is a common benign salivary gland neoplasm which is characterised by neoplastic proliferation of parenchymatous glandular cells along with myoepithelial components. It is the most common type of salivary gland tumor and most commonly affects the parotid gland. It derives its name from the architectural pleomorphism (variable appearance) seen by light microscopy. We report a case of Pleomorphic adenoma of the upper labial mucosa in a 66 -year-old male patient who reported with a swelling of the maxillary labial mucosa. A subsequent biopsy and immunohistochemistry were instrumental in this diagnosis. What makes this case significant is that the

incidence of primary PA in the minor salivary glands of the upper lip which is not very commonly specified earlier in the literature.

Keywords: Biopsy, Lip, Pleomorphic adenoma and Salivary gland tumors.

Introduction

The most common salivary gland tumor is Pleomorphic adenoma (PA), which accounts for 60–65% of such diseases. According to 2005 World Health Organization classification of salivary gland tumors, PA is an epithelial type tumor that is benign. On the basis of a population-based study in the United States, PA is extremely rare with an estimated annual age-adjusted incidence varying from 1 in 30,000 to 50,000 persons.¹

PA is the most common tumor of the salivary gland and accounts for 70-80 % of all benign tumors of the major salivary glands.² The name “Pleomorphic Adenoma” was suggested by Willis due to its unique histopathology characteristics.³ Pleomorphic adenoma is a morphologically complex entity as the epithelial and myoepithelial tumor cells can differentiate into fibrous, hyalinized, mucoid, myxoid, chondroid, osseous, or lipomatous tissue. Classically, PA is an encapsulated tumor; with nonmalignant lateral extensions into the capsule commonly encountered.⁴ The lesion most often arises in the superficial lobe of the parotid gland but may also be seen in the submandibular and minor salivary glands.⁵ Of all the pleomorphic adenomas over 85% occur in the parotid gland, 8% occur in the submandibular gland, and 6.5% occur in the minor salivary glands and 0.5 % in the sublingual glands. The submandibular gland is involved in only 5% to 10% of the salivary gland tumors, and pleomorphic adenoma (PA) is the most common tumor affecting it. Tumors arising in the minor salivary gland account for 22% of all salivary gland neoplasm. Most of them are malignant out of which 18% are benign.

Case Report

A 66-year-old male patient was referred to the Department of Oral Surgery with a diffuse swelling involving the maxillary mucosa. The swelling was insidious in onset and gradually increased in size. There was asymmetry of the face due to the swelling, and no altered sensation. On intraoral inspection the patient was edentulous and a solitary diffused swelling was evident on the maxillary labial mucosa. Extension of the soft tissue swelling was in the region of maxillary right lateral incisor till the first premolar. Swelling appeared pinkish white in colour with a smooth surface. On palpation there was no local raise in temperature and the swelling was non tender. It was firm to hard in consistency and the surface was smooth [Figure

1]. Extraorally overlying skin was easily pinchable. The patient on general examination looked alert and oriented, revealed normal gait, temperature, respiratory rate and pulse. Intra oral radiograph did not prove to be helpful. Blood investigations revealed that all parameters were within normal limits. A provisional diagnosis of traumatic fibroma, lipoma, and benign minor salivary gland tumour were considered. Excisional biopsy was carried out after which the biopsy specimen was processed and haematoxylin and eosin stained sections revealed an encapsulated tumor showing a fibrous capsule [Figure 2], Fluid filled ductal structures [Figure 3] were quite evident. The epithelium is seen forming ducts and cystic structures [Figure 4], areas depicting normal salivary gland tissue, a mixture of glandular epithelium and myoepithelial cells within a mesenchyme like background, also few areas show islands or sheets of cells [Figure 5]. Hyalinised, cartilaginous, bone [Figure 6 a] and myxoid [Figure 6 b] like tissue were also appreciated. All these features were suggestive of pleomorphic adenoma. A special stain PAS was done to check and confirm [Figure 7].

Discussion

Pleomorphic adenoma (PA) from the benign tumors is the most common one. The most common site of a PA of the minor salivary gland is the palate followed by lip, buccal mucosa, floor of mouth, tongue, tonsil, pharynx, retromolar area and nasal cavity.⁶

Pleomorphic adenomas are usually seen in middle aged women and present as a painless slowly growing mass. The lesions are usually solitary, ovoid, well demarcated masses. The larger tumors may have pedunculated outgrowths from the main lesion that grossly simulate multiple masses.² Signs and symptoms vary according to the location and size of tumor. Typically, PA presents as a slowly growing, mobile, and discrete nodule. Patients are often asymptomatic and the lesion is an incidental finding

during unrelated medical or dental visit, as seen in our patient. As the tumor grows, later findings include ulceration, pain, paresthesia, dysphagia, speech impairment, referred otalgia, or even a rarely facial paralysis secondary to extrinsic compression of the seventh cranial nerve.⁷ Pleomorphic adenoma is more often seen in women in the fourth to sixth decade of their life, and the etiology is unknown.⁸ Physical examination alone does not suffice in distinguishing benign tumors of minor salivary gland from malignant types. Therefore a biopsy is indicated, which should be performed to ensure the lesion is benign. Preoperative imaging, incisional or excisional biopsy, or fine-needle aspiration biopsy must be done prior to surgery.⁶ In our case, due to relatively small size of the tumor, excisional biopsy was performed to confirm the diagnosis of a benign epithelioid tumor, that is, PA.

The criterion standard for treatment of PA is complete excision with an adequate margin. Incomplete resection of all minor salivary gland tumors leads to recurrence and yields difficulty in future operations. Pleomorphic adenoma relapse is estimated to occur in 5% to 30% of cases and is almost always a result of incomplete surgical resection that especially manifests in the form of multiple foci and may be found to be aggressive.⁹ Pleomorphic adenoma also carries a high rate of implantability; therefore, caution must be exercised not to rupture the capsule or leave residual tumor cells behind including the extensions into the surrounding tissues.¹⁰ Carcinoma ex-PA arises from untreated PA and occurs up to 3% of minor salivary gland tumors. The prognosis of PA after complete excision is excellent.

Conclusion

Pleomorphic adenomas are frequently asymptomatic benign lesions, patients may not be aware of their existence, or the tumor is discovered incidentally in many

cases. Rarely may they present in the minor salivary glands with unusual location, that is, the lower lip, as in the presented case. The treatment of choice after an appropriate workup is a complete excision. Follow-up at least after 5 years is warranted to rule out recurrence.

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Legend Figures



Figure 1: Intra oral view and gross pathology

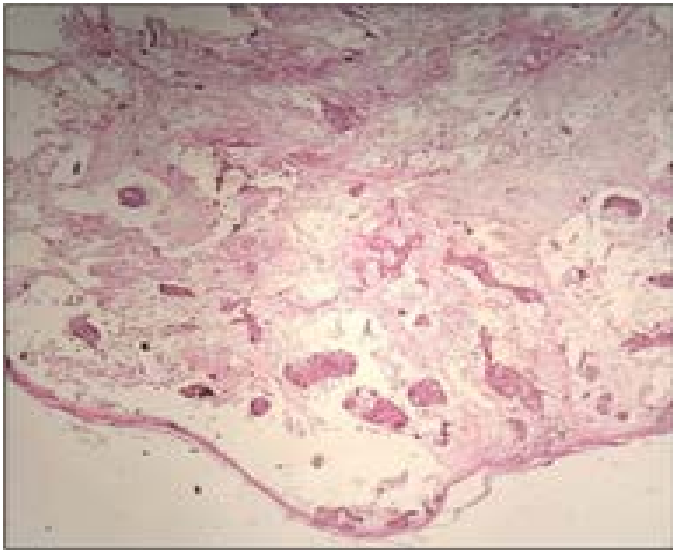


Figure 2: H & E stained section (4x magnification) showing the capsulated tumour mass.

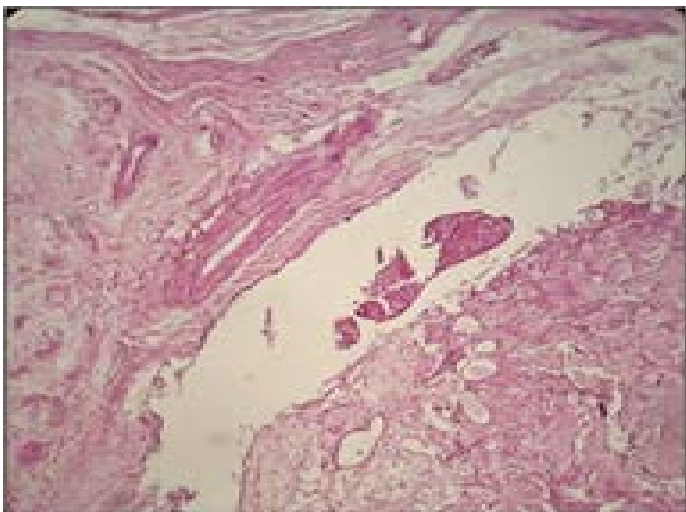


Figure 3: H & E stained section (10x magnification) showing the connective tissue capsule & fluid filled ductal structures.

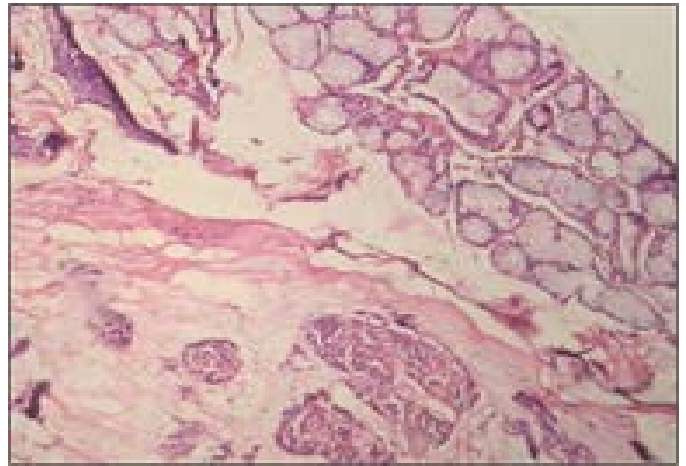


Figure 4 : H&E stained section (40x magnification) showing the normal salivary gland tissue.

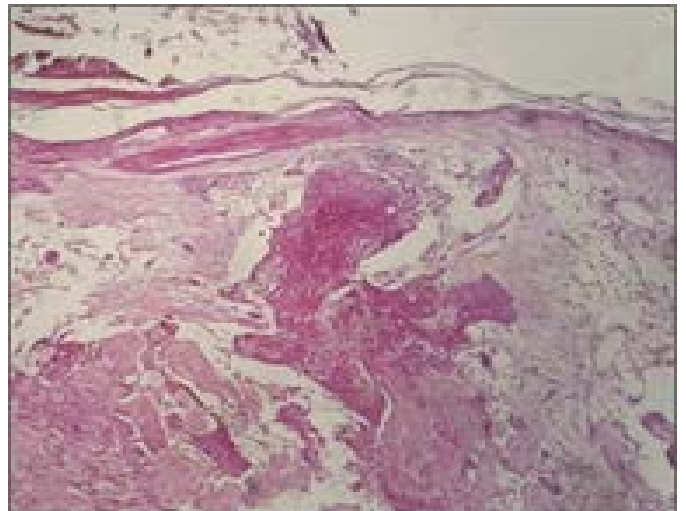


Figure 5: H& E stained section (40x magnification) showing the epithelial component & myxoid stroma.

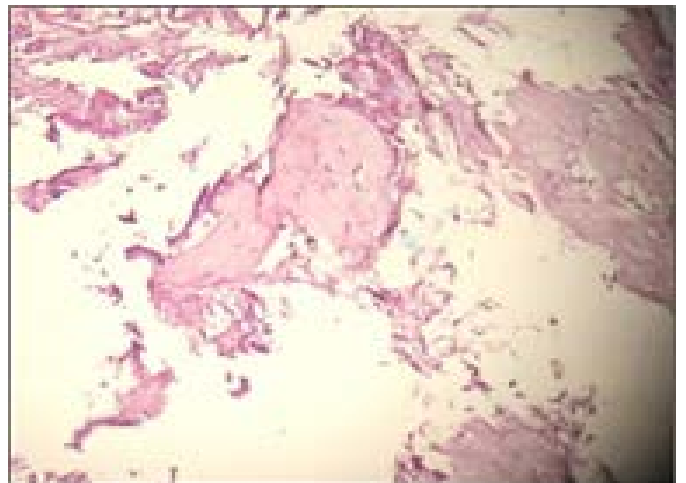


Figure 6(a): H & E stained section (40x magnification) showing the osseous tissue.

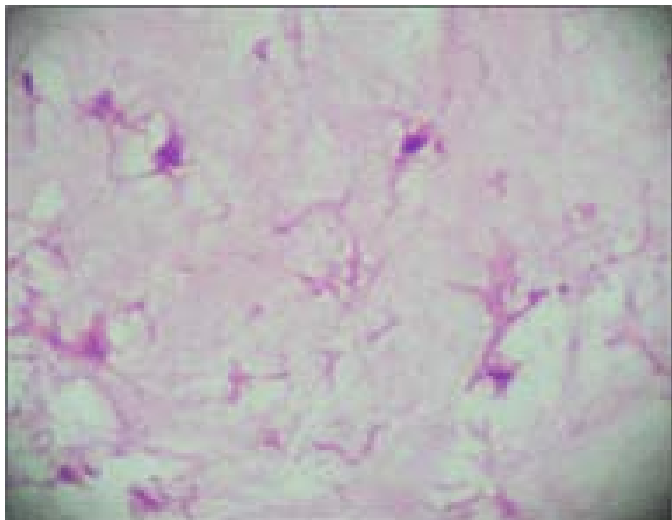


Figure 6(b): H & E stained section (40x magnification) showing the myxoid tissue.

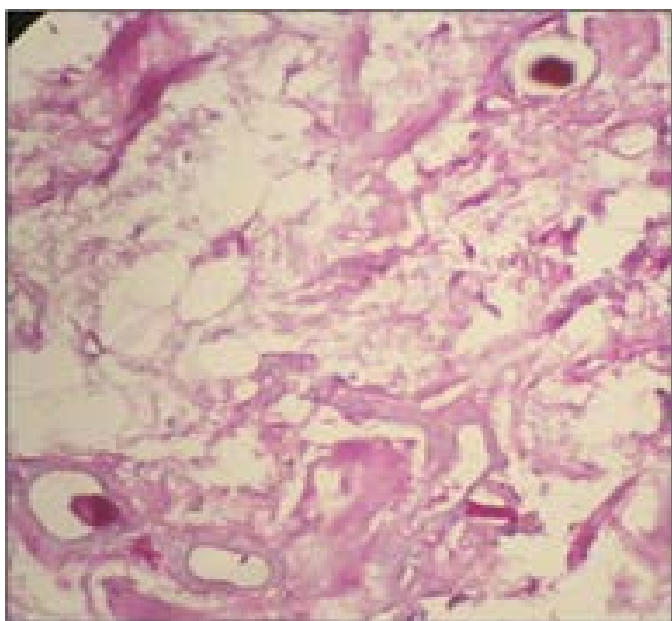


Figure 7: PAS stained section (40x magnification) showing the PAS +ve fluid in the ducts.