



Case Report

Upper Lip Submucosal Swelling – A Case Report

A. Fazeela Farzana^{1*}, Kirty R. Nandimath², Venkatesh G. Naikmasur³, Krishna N. Burde⁴, Kaveri Hallikeri⁵

¹Postgraduate Student, ^{2&3}Professors, ⁴Professor & Head, Department of Oral Medicine and Radiology, SDM College of Dental Sciences and Hospital, Dharwad, Karnataka, India.

⁵Professor & Head, Department of Oral Pathology and Microbiology, SDM College of Dental Sciences and Hospital, Dharwad, Karnataka, India.

ABSTRACT

Pleomorphic adenoma, also called benign mixed tumor, is the most common tumor of the salivary glands. Most of the Pleomorphic adenoma occurs at the parotid gland (85%) and the submandibular gland (8%). Intraoral Pleomorphic adenoma accounts for only 7%, which commonly involve the palate, uncommonly upper lip or buccal mucosa. This case report describes a rare and unusual lesion found in an older female, which was diagnosed as pleomorphic adenoma of the minor salivary glands in the upper lip. The tumor was a circumscribed, submucosal nodule, about 3x2cm in diameter and characterized by slow growth and firm in consistency. Complete excision was performed and the histopathological analysis showed an epithelial salivary gland tumor with islands of plasmacytoid cells, duct-like structures, in a variable stroma with chondroid, fibrous and myxoid appearance. The tumor did not recur. A brief review of the relevant literature is also presented.

KEYWORDS: Benign salivary gland tumour; Pleomorphic adenoma; Salivary gland

INTRODUCTION

Salivary gland tumours are quite rare and constitute 2%-6.5% of all head and neck tumours[1]. Minor salivary gland tumours accounts for 22% of all salivary gland neoplasms. Majority of them are malignant with only 18% being benign, of which commonest is pleomorphic adenoma(PA)[2]. They mostly arise in the parotid or submandibular salivary gland and the minor salivary glands in the oral cavity.

Minor salivary glands are located in palate, gingiva, floor of mouth, upper and lower lips, cheek, tongue, tonsillar areas, nasal cavity, paranasal sinuses, ears, jaw, pharynx, larynx, trachea & bronchi - may give rise to inflammatory conditions, benign & malignant tumors. The palate is the most common intraoral site (42.8%–68.8%), then upper lip (10.1%) and cheek (5.5%). Other rare sites include the throat (2.5%), retromolar region (0.7%), floor of the mouth and the alveolar mucosa[3]. Some of these pleomorphic adenoma can become massive with malignant degeneration before presentation.

Dardick and his associates stated the role of ductal reserve and myoepithelial cells in the histogenesis of pleomorphic adenoma. It is epithelial in origin, and clonal chromosome

abnormalities with aberrations involving 8q12 and 12q15 have been described[4]. Both PLAG1 and HMGA2 gene translocations have been identified as tumor-specific in pleomorphic adenoma, aids in establishing the diagnosis[5].

This case report describes the diagnosis and management of an asymptomatic, slowly growing, pleomorphic adenoma in the upper lip of an old aged female. A brief review of the relevant literatures is also presented.

CASE REPORT

A 60 years female patient reported with painless submucosal swelling in left half of the upper lip since 2years to the Department of Oral Medicine and Radiology, SDM College of Dental Sciences and Hospital, Dharwad, Karnataka. Patient noticed swelling which was less than 5mm, 2 years back since then it was gradually increased; it was asymptomatic with no history of discharge, bleeding, pain, or any sensory changes. Her past medical history was uneventful and there was no history of trauma. On extra oral examination, there is evidence of obliteration of left nasolabial fold. On intra oral examination the submucosal

swelling was indurated with 3x2cm in diameter, well circumscribed, superficial, mobile in all planes, non tender, firm in consistency, and overlying mucosal surface was smooth with a pinkish color showing evidence of superficial vascularity (**Figure 1**). There was no regional lymphadenopathy and her general conditions were within normal limits. Thus provisional diagnosis was given as benign non-odontogenic tumor especially considering the benign salivary gland tumors like pleomorphic and basal cell adenoma. Other considerations were tumors of neural origin namely encapsulated neurofibroma and schwannoma. Fine needle aspiration cytology of mass was performed, which was negative. After obtaining an informed consent from the patient, the entire tumor was excised with wide margin under local anaesthesia and sent for histopathological examination.

Figure 1: Intra oral view showing, non-ulcerative, submucosal swelling in the left half of the upper lip, overlying normal mucosa with superficial vascularity.



Gross specimen appeared solid ovoid, well encapsulated mass with multinodular appearance. The cut surface of the lesion was yellowish hue in colour (**Figure 2**). Histologically, showed capsulated tumour mass which consist of epithelial and mesenchymal component. Tumour is highly cellular and these epithelial cells are seen arranged in the form of bilayered ducts, strands, groups and sheets. Cells lining the duct are cuboidal with eosinophilic material in the lumen (**Figure 3A**).

Areas of hyalinization around the epithelial cells, myxoid areas with stellate shaped cells and chondroid like areas are evident in between the epithelial component (**Figure 3B**). Tumour cells are seen within capsule (**Figure 3C**). These features were consistent with pleomorphic adenoma of the minor salivary glands involving the upper lip. The wound healed uneventfully (**Figure 4**) and subsequent follow up after one year showed no signs of recurrence.

Figure 2: Gross specimen showed well encapsulated mass with multinodular appearance. Cut section showed yellowish hue.



Figure 3: 3A-Photomicrograph showed the epithelial and mesenchymal components.[H&E 4X] 3B-Background stroma composed of myxoid and chondroid areas.[H&E 10X] 3C-well defined capsule with tumour cell infiltration.[H&E 10X]

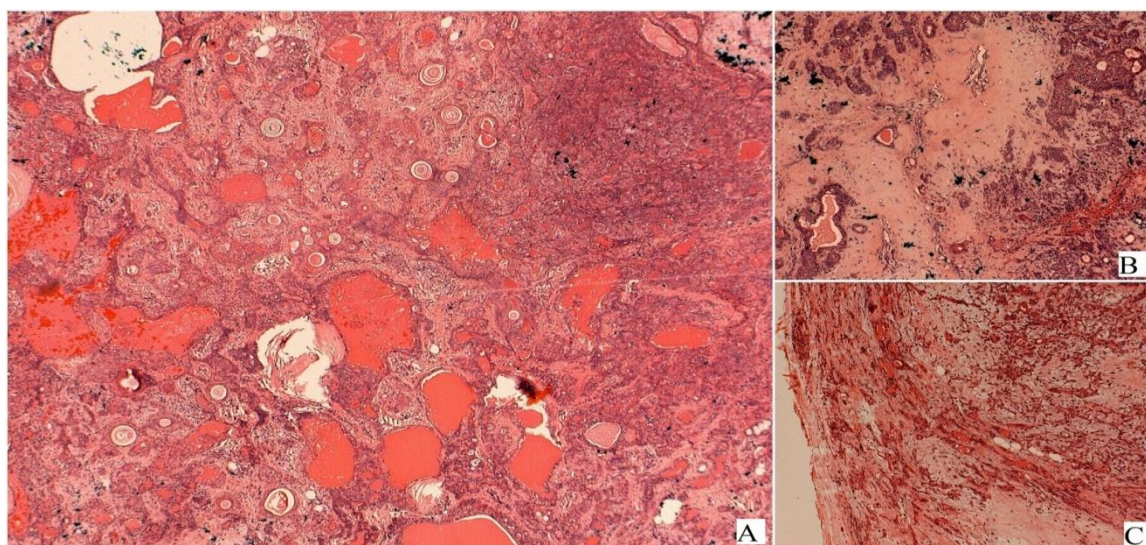


Figure 4: Post-operative photo after 3 weeks



DISCUSSION

Pleomorphic adenoma is the most common benign mixed tumor of salivary gland origin. Amongst the minor salivary glands most common site is hard & soft palate, the lip is relatively uncommon[6]. Minor salivary glands, are widely distributed in the head and neck region, approximately 450-1,000 in number. In many larger series of minor salivary gland tumors had reported a gender distribution of 66% in women; similarly the present case was reported in a female patient. The most frequent site of origin is in the hard palate because of the highest density of minor salivary glands. Most of the patients present a painless, non ulcerative, submucosal swelling. The mucosal layer is adherent to the mass and a small ulcer may be present during its evolution and 26% of patients may report with local pain[7].

Pleomorphic adenoma arising from minor salivary glands of the lips tends to occur at an earlier age than it does at other sites. Bernier found that the peak incidence of pleomorphic adenoma of the lips was in 3rd to 4th decade of life, with an average age of 33.2 years[8], but in this case report, patient's age was 60 years.

According to Pires FR et al[9], minor salivary gland tumors represent 9%-23% of all salivary gland tumors. 4042 cases of pleomorphic adenoma of the salivary glands were reviewed by Kroll and Hick. Of these, 445 originated in the minor salivary glands, of which only 16.9% in the upper lip and 2.9% in lower lip[10]. A study on 75 patients with intraoral minor salivary gland tumors, in that 15 of the 29 benign tumors were palatal in location with only four on the upper lip & one on the lower lip has reported by Jaber[11].

The ratio of about 6:1, considering the occurrence of pleomorphic adenoma in the upper lip with that of lower lip. The reason for this difference has been thought to be due to the differences in embryonic development between the upper and lower lips. There is a propensity for benign tumor to occur mostly in the upper lip, whereas malignant lesions tends to predominate in the lower lip[12].

Owens and Calcaterra[13], found 90% of the upper lip tumors to be benign in their reports. In the study by Neville

et al[14], 92% of the upper lip tumors were monomorphic adenoma (canalicular and basal cell adenoma) and pleomorphic adenoma. Owens and Calcaterra[13], found that 7 of the 13 malignant tumors in the lower lip were mucoepidermoid carcinoma. This finding was also consistent with the report from Neville et al[14], which confirmed 80% of lower lip tumors are composed by mucoepidermoid carcinoma.

However, suspicion of malignancy necessitates a biopsy before surgical treatment. Histologically, it is characterized by a large variety of tissues consisting of epithelial cells arranged in a cord-like cell pattern, together with areas of squamous differentiation or with plasmacytoid appearance myoepithelial cells which are responsible for the production of abundant extracellular matrix with chondroid, collagenous, mucoid and osseous stroma[15].

For most patients with benign, well-circumscribed tumors, the prognosis is excellent after surgical resection. However, recurrence can be a problem with pleomorphic adenoma and has been related to many factors, including an incomplete capsule, microscopic satellite tumor nodules, pseudopodia, capsular penetration may be seen beyond the capsule, and intraoperative tumor rupture, in which tumor contents spill into the operative field[16], in addition to those cases were treated with simple excision or surgical resection was performed with inadequate surgical margins. Multinodular tumor pattern is characteristic in recurrent pleomorphic adenomas.

Spiro reported a recurrence in 7% of 1342 patients with benign parotid neoplasms and 6% of patients with benign minor salivary gland tumours[17]. However, risk for recurrence can be expected in this patient, because of evidence of tumour cells seen within the capsule and high mesenchymal content particularly chondroid and myxoid stroma. Hence subsequent follow up was done till one year, revealed no recurrence. The surgical treatment for PA is a complete wide surgical excision with a good safety margin.

CONCLUSION

Pleomorphic adenoma of the lip is a rare neoplasm and therefore its diagnosis requires a high index of suspicion. Recurrence after surgical excision as well as malignant transformation should be a concern in this case report and therefore long-term follow-up is necessary. In India a rare conversion of pleomorphic adenoma to carcinoma has been reported, this makes the early diagnosis of such seemingly benign lesions even more urgent.

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REFERENCES

1. Kucuk U, Tan S. Pleomorphic adenoma of the upper lip. *Turk Patoloji Derg* 2011;27:73-6.
2. Varghese BT, Sebastian P, Abraham EK, Mathews A. Pleomorphic adenoma of minor salivary gland in the parapharyngeal space. *World J Surg Oncol* 2003;1(1):2.
3. Dalati T, Hussein MR. Juvenile pleomorphic adenoma of the cheek: a case report and review of literature. *Diagn Pathol* 2009;4:32.
4. Rajendran R. Tumors of the salivary glands. In: Rajendran R, Sivapathasundaram B, editors. *Shafer's textbook of oral pathology*. 6th ed. Noida: Elsevier; 2009. pp. 221.
5. Matsuyama A, Hisaoka M, Nagao Y, Hashimoto H. Aberrant PLAG1 expression in pleomorphic adenomas of the salivary gland: a molecular genetic and immunohistochemical study. *Virchows Arch* 2011;458(5):583-92.
6. Debnath SC, Adhyapok AK. Pleomorphic adenoma (Benign mixed tumor) of the minor salivary glands of the upper lip. *J Maxillofac Oral sur* 2010;9(2):205-8.
7. Vander Poorten VL, Balm AJ, Hilgers FJ, Tan IB, Keus RB, Hart AA. Stage as major long-term outcome predictor in minor salivary gland carcinoma. *Cancer* 2000;89(6): 1195-204.
8. Vernier JL. Mixed tumors of lips. *J Oral Surg (Chic)* 1946;4:193-202.
9. Pires FR, Pringle GA, de Almeida OP, Chen SY. Intra oral minor salivary gland tumors: a clinicopathological study of 546 cases. *Oral Oncol* 2007;43(5):463-70.
10. Krolls SO, Hicks JL. Mixed tumors of the lower lip. *Oral Surg Oral Med Oral Pathol* 1973;35(2):212-7.
11. Jaber MA. Intraoral minor salivary gland tumors: a review of 75 cases in Libyan Population. *Int J Oral Maxillofac Surg* 2006;35:150-4.
12. Shrestha A, Reddy NS, Ganguly SN. Pleomorphic adenoma of the upper lip: A case report. *Nepal Med Coll J I* 2010;6(1):51-3.
13. Owens OT, Calcaterra TC. Salivary gland tumors of the lip. *Arch Otolaryngol* 1982;108(1):45-7.
14. Neville BW, Damm DD, Weir JC, Fantasia JE. Labial salivary gland tumors. *Cancer* 1988;61(10):2113-6.
15. Lotufo MA, Junior CA, Mattos JP, Franca CM. Pleomorphic adenoma of the upper lip in a child. *J Oral Sci* 2008;50(2):225-8.
16. Riad MA, Abdel-Rahman H, Ezzat WF, Adly A, Dessouky O, Shehata M. Variables related to recurrence of pleomorphic adenomas: outcome of parotid surgery in 182 cases. *Laryngoscope* 2011;121(7):1467-72.
17. Spiro RH. Salivary Neoplasms: overview of a 35 year experience with 2807 patients. *Head Neck Surg* 1986;8(3):177-184.

*Corresponding author: Dr. A. Fazeela Farzana
E-Mail: dr.fazeela86@gmail.com