



PLEOMORPHIC ADENOMA OF LIP : A RARE CASE REPORT AND REVIEW OF LITERATURE

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ABSTRACT

Minor salivary gland tumors are rare, comprise 2-5% of head and neck tumors. The palate is the most common site for oral minor salivary gland tumors. Pleomorphic adenoma, the most common salivary gland tumor, is also the most common benign salivary gland tumor. Lip is infrequently involved by pleomorphic adenoma while canalicular adenoma shows exclusive minor salivary gland manifestation with a higher predilection toward the upper lip. Considering the clinical similarity with canalicular adenoma, incomplete encapsulation, diverse microscopic patterns, histopathological features associated with recurrence, and potential for malignant transformation; pleomorphic adenoma demands both diagnostic and surgical expertise for accurate diagnosis and optimal surgical outcome.

KEYWORDS : Minor salivary gland tumors, pleomorphic adenoma, canalicular adenoma, pseudoencapsulation

INTRODUCTION

Minor salivary gland tumors account for only 2–5% of all head and neck tumors and of them approximately 70% occur in the oral cavity, 25% in the nasal cavity/sinuses/nasopharynx and 3% occur in the larynx.¹ Of the oral minor salivary gland tumors, at least 50% have been diagnosed in the palate, representing the most common site for oral minor salivary gland tumors.^{2,3} Occurrence on the lips appears to account for approximately 5% of these tumors, with most occurring on the upper lip. The upper lip is ten times more likely to be involved than the lower lip.⁴

Relative frequency of benign versus malignant tumors involving minor salivary glands has varied widely among different studies, and can also be found in two of the largest series on minor salivary gland tumors. Spiro³ reported a higher frequency of malignant tumors (87%) in contrast to Auclair PL,⁵ with reporting frequency of malignant tumors at 49%. A large difference in the sample size (607 versus 2945) might have resulted in this contrasting difference. Yih WY⁶ et al also reported benign intraoral minor salivary gland tumors (56%) to be more common than malignant minor salivary gland tumors (44%). In most Indian studies, malignant tumors were found to involve minor salivary glands more frequently as compared to benign tumors.^{7,8,9} Subhashraj K⁸ however reported a very small difference in benign (22%) and malignant (23%) tumors involving minor salivary glands. Laishram RS et al¹⁰ found an equal incidence of benign and malignant tumors of minor salivary glands. Mishra S et al¹¹ however found benign tumors to be involving minor salivary glands more frequently when compared to malignant tumors.

Pleomorphic adenoma is the most common salivary gland tumor. A study by Auclair PL⁵ reporting 13,749 salivary gland tumors showed 6880 cases of pleomorphic adenoma of which 4359 were located in the parotid gland and 1277 were located in minor salivary gland tissue. Palate accounted for 711 of these 6880 cases of pleomorphic adenoma (10.3%) representing the second most common site after parotid and 56% of the cases located in the minor salivary glands. Yih WY⁶ also reported pleomorphic adenoma as the most common benign tumor (78.2%) followed by canalicular adenoma and

mucoepidermoid carcinoma as the most common malignant tumor (47.9%) followed by adenoid cystic carcinoma. The palate was the most common site of involvement in their study. Similarly most of the Indian series have also reported pleomorphic adenoma to be the most common benign tumor in minor salivary gland with the palate as the predilected site and mucoepidermoid carcinoma as the most common malignant minor salivary gland tumor. In contrast, Vani NV et al⁹ reported mucoepidermoid carcinoma to be the most common tumor followed by pleomorphic adenoma while Subhashraj K⁸ found adenoid cystic carcinoma as the most common malignant minor salivary gland tumor.

CASE REPORT

A 78 year old female reported to the department with the chief complaint of a mass on the left side of the upper lip from last one and half years. Mass was initially smaller in size and gradually increased over the initial 5-6 months to attain present dimensions of approximately 7mm in diameter after which the growth ceased. Patient reported no spontaneous signs and symptoms associated with it except occasional pain and that too on palpating it firmly.

Patient gave the history of multiple tooth extractions 4 years back, after which she got her maxillary and mandibular dentures fabricated and had been using them comfortably since then until recently when she started feeling a mass in the upper lip while talking and chewing food. Medical, drug, social, family, and personal history were non-contributory.

On inspection, there was no gross extraoral swelling while a well-defined and localized swelling with normal overlying mucosa was seen intraorally involving upper labial mucosa on the left side. On palpation, a well-defined, smooth-surfaced, submucosal, non-tender, non-fluctuant, firm, and freely movable mass was palpable. Mass was round, approximately 7-8 mm in diameter, and with a normal temperature of overlying mucosa. No lymphadenopathy was detected on palpation.

Based on the history and clinical examination, various differentials which could be assigned to this lesion were – mucocele, canalicular adenoma, basal cell adenoma,

pleomorphic adenoma, fibroma, lipoma, and sialolithiasis. Radiographic examination for the mass revealed no significant findings and ruled out the calculus of the labial minor salivary gland. Furthermore pain and inflammation are common with sialolithiasis.¹² Mucocele and fibroma follow some irritational or traumatic incident which is more common with the lower lip. Trauma from ill-fitting denture was ruled out in our case as the patient didn't report any trauma / problem associated with the denture wearing. Also, fibromas are most often sessile or slightly pedunculated and swelling with mucocele is soft, fluctuant, and shows characteristic fluctuation in its size, appearing suddenly and reaching maximum size within several days. Whereas superficial mucoceles have slightly bluish / translucent appearance, deeper lesions may have normal colour. Lipomas also are soft and sometimes almost fluctuant to palpation.¹³ Basal cell adenoma and pleomorphic adenoma have parotid as the predilected site with more than 70% of them appearing as parotid masses. At oral mucosal sites basal cell adenoma shows a predilection for the upper lip while pleomorphic adenoma shows a predilection for the palate. Lip is rarely involved by the pleomorphic adenoma. Basal cell adenoma is more common in males with an average age of 60 years and ranges between 1 – 3 cm in size. Pleomorphic adenoma is more common in females with an age range of 30 – 50 years and is larger in size.^{12,14}

Canalicular adenoma occurs exclusively in minor salivary glands with approximately 80% of cases involving the upper lip and near the midline and presenting as painless freely movable mass in the upper lip submucosa. This tumor is much more common in females (older than 50 years) as compared to males and ranges in sizes between 0.5 – 3cm.^{13,14}

Based on the above discussion, a provisional diagnosis of canalicular adenoma was assigned to the lesion. Considering the location, clinical features and, size of the lesion no preoperative incisional or fine-needle aspiration biopsy was performed and excision of the mass was planned for both diagnostic and therapeutic purposes.

The lesion was excised under local anesthesia by placing an incision over mass and then performing gentle and careful blunt dissection. The specimen was sent for histopathological examination.

On histopathological examination, an encapsulated tumor (round or angular in shape) was found with salivary gland epithelium proliferating inside in the form of duct and sheets. Ducts were variable in size and contained eosinophilic coagulum. Histopathological report revealed the features consistent with histological features of pleomorphic adenoma.

Patient was followed at 3 days and 1 week postoperatively. Post-operative course was uneventful and healing was found to be satisfactory. Patient was maintained on regular follow up and no recurrence was found until recently at approximately three and half years postoperatively.

DISCUSSION

Salivary gland neoplasms other than pleomorphic adenoma and canalicular adenoma are rare in the upper lip. Pleomorphic adenomas are firm, nodular growths, which generally occur in younger patients than the smooth-surfaced, sometimes fluctuant canalicular adenoma.¹²

A canalicular adenoma is a benign tumor that has a significant predilection for the upper lip. The canalicular adenoma classically occurs in the upper lip in elderly women.¹⁴ Of the total 121 canalicular adenomas reported by Auclair PL,⁵ 89 occurred in the upper lip followed by buccal

mucosa as the second most common site. Canalicular adenomas typically affect an older population compared to pleomorphic adenomas.¹⁵ Canalicular adenoma is typically an asymptomatic, slow-growing, and freely movable mass that uncommonly exceeds 2 cm in widest diameter. About 20% will occur multifocally, requiring a complete examination of the entire lip for smaller, developing tumors.¹⁴

Treatment for pleomorphic adenoma of lip consists of excision of the mass with 1-cm margins, while for canalicular adenoma includes excision with 0.5-cm margins. Excision should include the overlying epithelium and the muscle fascia of the orbicularis oris muscle. Enucleation, or a "shelling out" of a pleomorphic adenoma, is contraindicated. The pseudo capsule of a pleomorphic adenoma will certainly give the clinical impression of complete removal of an "encapsulated nodule or mass" with these approaches, but the extracapsular tumor projections left behind may lead not just to recurrence but to multicentric recurrences caused by the remaining tumor projections and foci within the tissue at the circumference of the resections.¹⁴ Simple enucleation of canalicular adenoma is, apparently, curative, despite occasional intracapsular foci of the tumor, while similar treatment of pleomorphic adenoma invites recurrence. Recurrence of canalicular adenoma has not been conclusively proven and multifocal lesions are reported in approximately 22% of the cases.¹² Multiple case reports however have reported enucleation as a treatment modality for both canalicular adenoma as well as pleomorphic adenoma. Stramandinoli-Zanicotti RT et al¹⁶ and Bahr JA¹⁷ et al carried out enucleation for canalicular adenoma and reported no recurrence at 8 month follow up. Similarly E Tucci¹⁸ also carried out enucleation of pleomorphic adenoma of the palate and reported no recurrence at 2 years follow up. Afonso RA et al¹⁹ reported a case where they carried out conservative shelling out of tumors by making and incision overlying the lesion and preventing rupture of the tumor capsule followed by complete tissue divulsion. Ahmedi JR et al²⁰ carried out complete excision of pleomorphic adenoma of upper lip by carrying out by careful dissection of encapsulated mass and without including soft tissue margins. No recurrence of the lesion was reported at 1 year follow up.

Although no recurrence was reported in all these studies but all these studies reported follow up period of relatively short duration. Recurrence with pleomorphic adenoma is seen at approximately 6 years following the initial surgery.¹⁴ Besides this, other factors associated with recurrence of pleomorphic adenoma include incomplete removal, incomplete capsulation, intraoperative spillage of tumor mucoid because of intraoperative burst of the tumor. Histopathological features linked to recurrence of pleomorphic adenoma include predominantly cellular or extremely cellular presentation, or hypocellular tumor in abundant myxoid or chondromyxoid stroma.²¹

The diverse microscopic pattern of pleomorphic adenoma is one of its most characteristic features.¹² Challenges to microscopic diagnosis also include biphasic differentiation of salivary gland tumors even to the point of hybrid tumors with features of two different, well-defined tumor entities. With few immunohistochemical markers available for differentiation of tumors, accurate diagnosis of minor salivary gland tumors may be quite difficult.

Pleomorphic adenomas are benign tumors with a well documented transformation to malignancy (carcinoma ex pleomorphic adenoma). It is estimated that up to 25% of untreated pleomorphic adenomas undergo malignant transformation, a process that is size and time related.¹⁴

Malignant transformation is suspected when the tumor grows rapidly, presents with pain or irregular borders or local

neurological disturbances such as anesthesia, paraesthesia, or it is covered by ulcerated/irregular mucosa or skin. Multiple recurrences also increases the risk of malignant transformation of pleomorphic adenoma.

CONCLUSION

Pleomorphic adenoma and canalicul adenoma represents two most common salivary gland tumors on lip. Simple enucleation of canalicul adenoma although is apparently curative, despite occasional intracapsular foci of tumor, similar treatment for pleomorphic adenoma may lead to recurrence. Considering the clinical similarity with canalicul adenoma, incomplete encapsulation, diverse microscopic patterns, histopathological features associated with recurrence and potential for malignant transformation; pleomorphic adenoma demands both diagnostic and surgical expertise for accurate diagnosis and optimal surgical outcome.

Surgical excision of the lesion with a cuff of apparently normal tissue may be warranted as a wise surgical option for both lesion. Although this procedure may constitute a slight over treatment of a canalicul adenoma, the increased morbidity is minimal, and the risk of recurrence from tumor within the capsule or from incompletely excised nonencapsulated lesions makes such treatment logical and defensible.

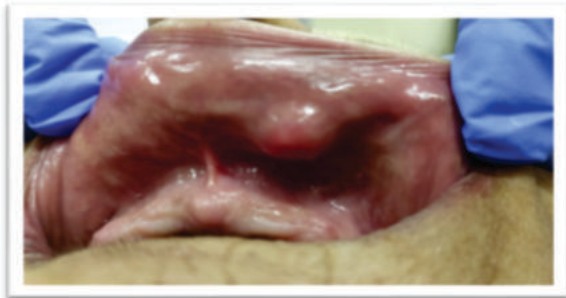


Figure 1. Clinical picture of the lesion intraorally



Figure 2. Surgical site after enucleation of lesion



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