

Pleomorphic adenoma of the upper lip: A rare case report

ABSTRACT

Pleomorphic adenoma (PA), also called mixed tumor, is the most common benign tumor of the salivary glands that mostly occurs in the parotid or submandibular glands but may also occur in the minor salivary glands that are distributed throughout the oral cavity. The common sites of PA of the minor salivary glands are the palates followed by lips and cheeks. Surgical removal with adequate margins is the principal treatment. We present a case of PA (arising from the minor salivary gland of the upper lip) – a rare site in a 65-year-old female who presented with a swelling upper lip of 2-year duration. Fine-needle aspiration cytology of the swelling revealed PA which was confirmed by histopathological examination after complete excision of the swelling.

Keywords: Minor salivary gland, pleomorphic adenoma, upper lip

INTRODUCTION

Pleomorphic adenoma (PA), or benign mixed tumor, is the most common salivary gland neoplasm, accounting for 60%–65% of all major and minor salivary gland tumors.^[1] Minor salivary glands are located in the palate, upper and lower lips, gingiva, floor of the mouth, cheek, tongue, tonsillar areas, nasal cavity, paranasal sinuses, ears, jaw, pharynx, larynx, trachea, and bronchi – where it may give rise to inflammatory conditions and benign and malignant tumors.^[2] Although the hard and soft palates are the most common sites of minor salivary gland tumors, the upper lip is relatively uncommon site. Eighty percent of the minor salivary gland tumors located in the lip are benign.^[3] It occurs frequently in females, with a female-male ratio ranging from 1.9:1 to 3.2:1 and with a peak incidence between the 5th and the 7th decades of life.^[4,5] This tumor presents as an asymptomatic firm mass with a long period of slow growth rate, whereas secondary to trauma, the clinical features may also include ulceration, pain, or bleeding.^[6] 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.^[7]

CASE REPORT


A 68-year-old female patient reported to our dental outpatient department with complaints of a painless swelling upper lip of 2-year duration which had gradually started increasing from the past 4 months. There was no antecedent history of trauma or any other medical history of significance. Extraoral examination revealed a swelling on the left side of the upper lip with normal overlying skin [Figure 1]. On palpation, the swelling was firm in consistency but nontender. On intraoral examination, a solitary well-circumscribed swelling about 2 cm × 2.5 cm, firm in consistency, mobile but nontender was noted in the upper lip on the left side of the midline. The overlying mucosa was smooth without any ulceration or toxicity

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[Figure 2]. The systemic examination was normal, and there was no regional lymphadenopathy. Routine baseline investigations were within normal limits. Fine-needle aspiration cytology (FNAC) of the swelling revealed epithelial components and myxoid stroma, suggesting the diagnosis of PA. Excision of the mass was planned under local anaesthesia [Figure 3 and 4]. Complete excision of the tumor was possible as the tumor was not fixed to the underlying structures, and a primary closure of the defect was done with good cosmetic result [Figures 5 and 6]. The histopathological section showed well-encapsulated soft-tissue mass consisting of epithelial and myoepithelial cell proliferation lining the ducts and chondromyxoid stroma with the final diagnosis of PA of the upper lip. The patient is on our follow-up without any signs of recurrence.

DISCUSSION

PA is the most common benign tumor of the minor salivary glands. Females are more affected than males. These tumors

are most often diagnosed in the 4th and 6th decades of life. The typical presentation is slow-growing painless firm mass and nontender and tends to be mobile when small but fixed to the surrounding tissue with advanced growth.^[8] Prognosis is good if the tumor is well excised. The most common sites among the major salivary gland are the parotid gland (approximately 75%), the submandibular gland (around 5%–10%), and the minor salivary gland (approximately 10%).^[9] Among the minor salivary glands, the most common sites are the hard palate and the soft palate, and the lip is relatively uncommon.^[10] The main etiopathogenesis of PA remains unclear. Cytogenetic and molecular studies have described that it is of epithelial origin with chromosomal abnormalities at 8q12 and 12q15.^[7,10]

PA of the lip is a rare neoplasm, and therefore, its diagnosis requires a high index of suspicion. It is an epithelial tumor of complex morphology, possessing epithelial and myoepithelial



Figure 1: Extraoral view of the swelling



Figure 2: Intraoral view of the swelling

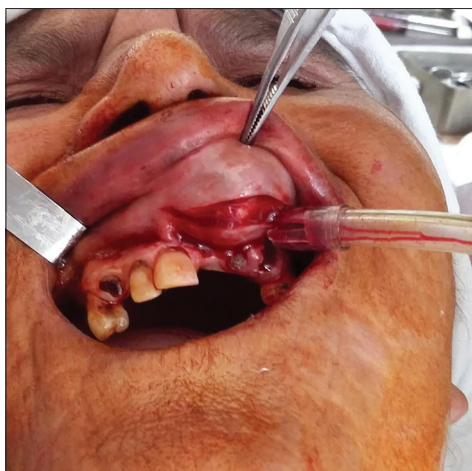


Figure 3: Intra Oral Mucosal Incision over the swelling



Figure 4: Excision of Swelling



Figure 5: Primary closure of the defect

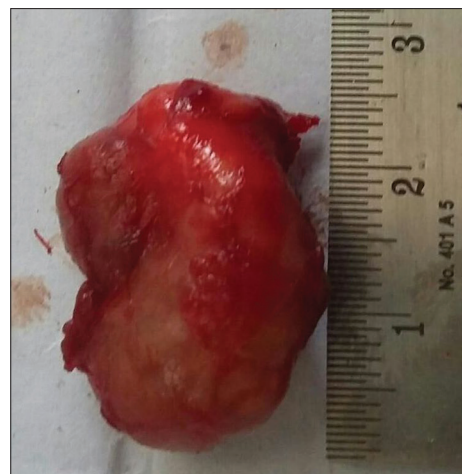


Figure 6: Excised specimen

elements arranged in varieties of patterns and embedded in mucopolysaccharide stroma.

A complete wide surgical excision is the treatment of choice. Recurrence after many years of surgical excision as well as malignant transformation should be a concern, and therefore, long-term follow-up is necessary.

CONCLUSION

PA in the minor salivary gland of the upper lip is very rare. FNAC is a valuable diagnostic adjuvant in preoperative evaluation of salivary gland lesions. It provides a preliminary diagnosis and preoperative assessment on which management decisions can be based. Although rare, PA should be considered as a differential diagnosis of swellings in the upper lip. This lesion, in most frequent cases, is asymptomatic, and the patient may not be aware of its existence and is discovered accidentally by a dentist. Although it is very difficult to diagnose the salivary gland diseases, histopathology is the only gold standard for diagnosis of all lesions.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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