Case Report

Pleomorphic Adenoma: Case Report and Review of Literature

N. Balan, M. Sudhaa Mani, S. Yasmeen Ahamed, D. A. Divya

Department of Oral Medicine and Radiology, Vivekanandha Dental College for Women, Namakkal, Tamil Nadu, India

Abstract

Pleomorphic adenoma, the most common salivary gland tumor, consists of epithelial and mesenchymal components. Its morphologic complexity results from differentiation of tumor cells into fibrous, hyalinized, myxoid, chondroid, and osseous areas. The diagnosis is made by the clinical and histopathological examination. The occurrence of pleomorphic adenoma in upper lip is very rare. Kroll and Hick reviewed 4042 cases of pleomorphic adenomas of the salivary glands. Of these, 445 originated in the minor salivary glands, of which only 16.9% were located in the upper lip and 2.9% in the lower lip. Pleomorphic adenomas of the minor salivary glands generally present as painless, submucosal swellings. We report a case of pleomorphic adenoma which presents as a swelling of upper lip for a 72-year-old female patient.

Keywords: *Minor salivary gland, mixed tumor, upper lip*

Introduction

The term pleomorphic adenoma was suggested by Willis. In earlier years, it was referred by names such as enclavoma, branchioma, endothelioma, and enchondroma.[1] It is the commonest of the salivary gland tumors, accounting for 50%–70% of cases of parotid tumors, 40%–60% submandibular tumors, and 10% minor salivary gland tumors, with palate (60%) being the most common followed by upper lip (20% of cases).[2] The most commonly affected age group are in the fourth, fifth, and sixth decades; 60% of them are female.[3] It has been suggested that 25% of benign mixed tumors undergo malignant transformation.

Case Report

A 72-year-old female patient reported to the Department of Oral Medicine and Radiology with the complaint of swelling in the left middle-third of face for the past 1 year. History revealed that the swelling was gradual in onset and slowly increased in size for the past 1 year. The swelling was asymptomatic. Patient reported of the nasal stiffness of the left nostril and associated difficulty in breathing. There was no history of trauma, loss of appetite, and loss of weight.

On extraoral examination, a well-defined swelling was present on left middle-third of the face, oval, 3 cm × 2 cm in size. It extended superiorly 3 cm from inner canthus of the eye, inferiorly to vermillion border of the upper lip, medially from infranasal depression laterally to nasolabial fold. There was deviation of the nasal septum to the right side, elevation of left ala of nose and obliteration of nasolabial fold. The surface appeared smooth with no secondary changes. On palpation, the swelling had no local rise in temperature, nontender, firm in consistency, and freely mobile. No pulsations were felt [Figure 1]. Bilateral submandibular lymph nodes were palpable, firm in consistency and mobile.

On intraoral examination, a well-defined swelling was present on the left maxillary labial mucosa, ovoid, 3 cm × 2 cm in size, surface was smooth and mucosa over the swelling appeared pale pink, extends anteriorly from maxillary labial frenum, posteriorly 1 cm away from buccal frenum, superiorly 0.5 cm from vermillion border of upper lip, inferiorly to attached gingiva. There were no secondary changes such as sinus opening or pus discharge or ulceration. On palpation, it was nontender, firm in

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Address for correspondence: Dr. M. Sudhaa Mani, Department of Oral Medicine and Radiology, Vivekanandha Dental College for Women, Tiruchengode, Namakkal, Tamil Nadu, India.
E-mail: drsudhaamanimds@gmail.com

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consistency, compressible, and mobile. No pulsations were felt [Figure 2]. Based on the history and clinical findings, a provisional diagnosis of benign salivary gland tumor was considered probably pleomorphic adenoma. The differential diagnosis was fibrosed mucocele, peripheral fibroma, myxoma, and lipoma. Fine-needle aspiration cytology was negative. Intraoral periapical radiograph and the OPG revealed no bony changes. CT report revealed a hypodense area of size 3 cm × 2 cm seen in relation to upper lip with mild scalloping of the underlying maxilla [Figure 3a and b]. Excisional biopsy was done under GA and the specimen was submitted for histopathological examination [Figure 4a and b] Microscopic analysis of the surgical specimen revealed an encapsulated mass with nests of epithelial cells in a background of myxoid stroma. These epithelial cells exhibit large nuclei with eosinophilic cytoplasm and indistinct cell borders. The surrounding myxoid stroma reveals widely separated angular shaped cells with numerous neurovascular bundles. The final diagnosis was pleomorphic adenoma [Figure 5a and b]. Follow up was done after 1 month and patient was free of symptoms [Figure 6].

Discussion

Pleomorphic adenoma is a benign neoplasm which consists of cells exhibiting the ability to differentiate into both epithelial (ductal and nonductal) cells and mesenchymal (chondroid, myxoid, and osseous) cells. The most common site among the major salivary gland is parotid (approximately 75%) submandibular gland (around 5%–10%) and the minor salivary gland (approximately 10%). In a study conducted in Indian population during 5 years duration, out of 5515 tumors involving various organs, 53 were salivary gland tumors accounting for 0.96% of all neoplasms.[4] Most cases of pleomorphic adenomas (70%) show cytogenetic aberrations.[5] The mucin 1 gene has been associated with malignant transformation of this tumor.[6] The literature suggests that the simian virus (SV40) may play a causative role in the

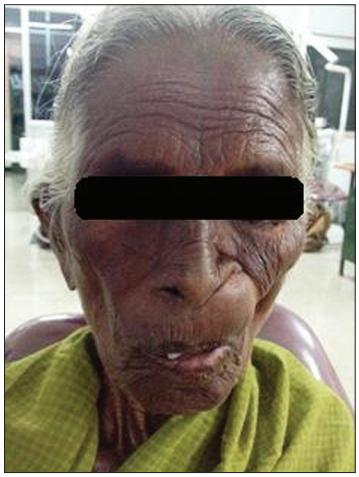

Figure 1: Extraoral photograph

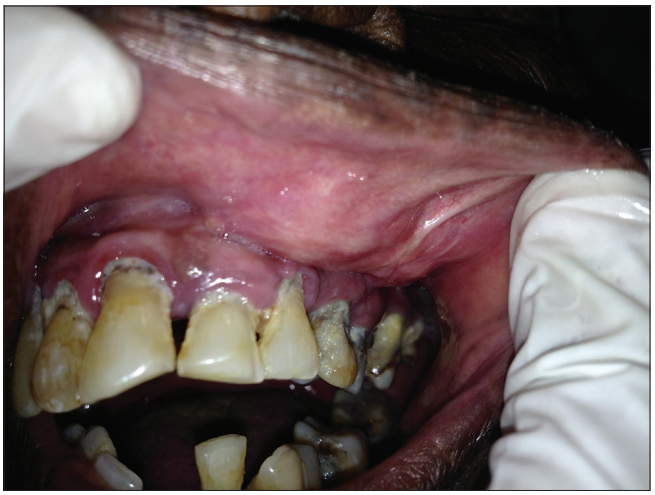

Figure 2: Intraoral preoperative view

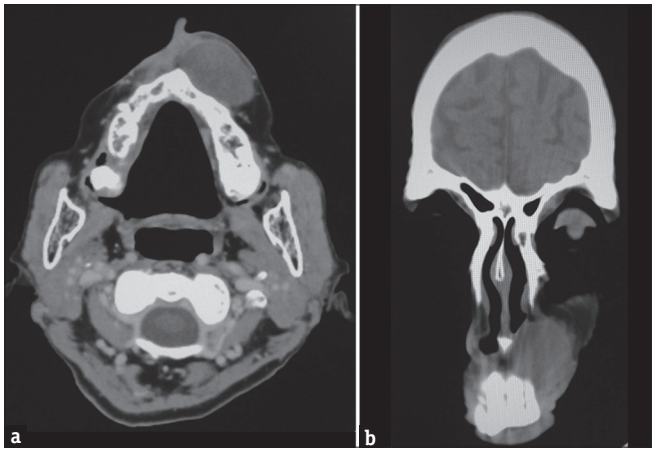

Figure 3: (a) Axial computed tomography scan image showing hypodense area in left premaxillary region. (b) Coronal computed tomography scan image showing hypodense area in left premaxillary region

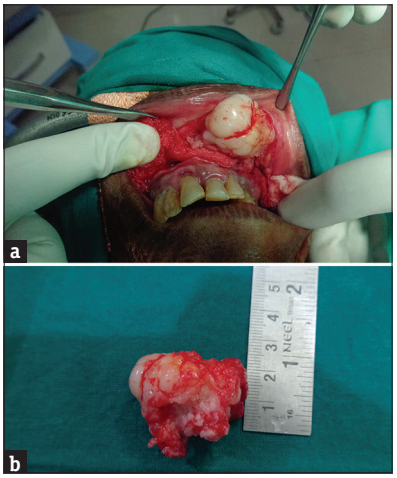

Figure 4: (a) Excision of lesion. (b) Measurement of the lesion

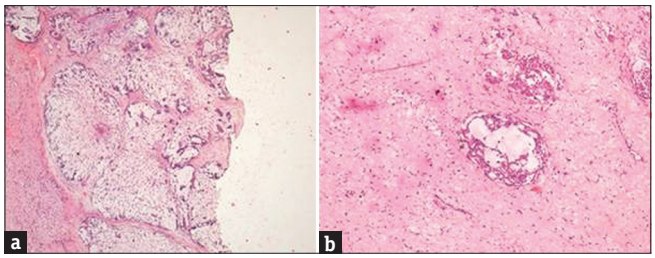

Figure 5: (a) Histopathologic view ×40. (b) Histopathologic view ×10

development of pleomorphic adenoma. Putative risk factors include cigarette smoking, genetic predisposition, viral infections, rubber manufacturing, plumbing, some types of woodworking, as well as asbestos mining, exposure to nickel compounds, and cellular phone use.[7] The only well-established risk factor is ionizing radiation. Atomic bomb survivors and cancer patients treated by radiation present with a substantially higher risk of developing salivary gland tumors. The typical presentation is asymptomatic, slow growing, painless, firm mass, nontender, and tends to be mobile when small but fixed to surround tissue with advanced growth.[8] In our case, the tumor presents as asymptomatic slow growing swelling which is nontender and firm in consistency and mobile. The extent and depth of the lesion cannot be accurately seen in conventional radiographs, henceforth the 3-d imaging modalities such as computed tomography (CT) scan and magnetic resonance imaging are considered to be gold standard in imaging such lesions. In our case, no bony changes seen in intraoral periapical and panoramic radiograph. On CT images, a hypodense area of size 3 cm × 2 cm seen in relation to upper lip with mild scalloping of the underlying maxilla. Radical surgical excision is the cornerstone treatment of salivary gland tumors.[9] If complete resection cannot be achieved, adjuvant radiotherapy should be added to improve local control.[10] The differential diagnosis of asymptomatic nodules involving minor salivary gland are neurofibroma, lipoma, and rhabdomyosarcoma.[11] In the study by Neville *et al*., 92% of the upper lip tumors were monomorphic adenoma (canalicular adenoma and basal cell adenoma) and pleomorphic adenoma, whereas sporadic cases of adenoid cystic carcinoma, acinic carcinoma, and adenocarcinoma constitute the remainder. Malignant tumors tend to predominate in the lower lip.[12] Most recurrences are due to inadequate surgical techniques such as simple enucleation leaving behind microscopic pseudopod-like extensions.[13]

Conclusion

Pleomorphic adenoma generally does not recur after adequate surgical excision. Ultimately, complete surgical excision will provide the definitive diagnosis and treatment for this noteworthy salivary gland neoplasm.

Figure 6: Postoperative view after 1 month

Prognosis is excellent as the tumor is benign, and recurrence is not being noted.

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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