Case Report

Pleomorphic adenoma of the upper lip

ABSTRACT

We present a rare case of pleomorphic adenoma in the upper lip region, with erosion of the maxillary bone and distortion of the facial appearance. A 20-year-old man presented with a painless mass on his upper lip, which had gradually increased in size over a period of 2 years. Computed tomography demonstrated a 30 mm × 28 mm enhancing mass in the upper lip region with no invasion to the surrounding tissues. Erosion of the maxillary bone posterior to this lesion was noted. The lesion was excised completely with a wedge of mucosa overlying the tumoral mass, accompanied with abdominal fat grafting to prevent labial asymmetry. Histological examination confirmed the diagnosis of a minor salivary gland pleomorphic adenoma in the upper lip. The pathology, clinical manifestations, and treatment of intraoral pleomorphic adenomas are reviewed.

Keywords: Lip, maxillary bone, pleomorphic adenoma

INTRODUCTION

Pleomorphic adenoma is frequently observed in the major salivary glands; however; the minor salivary glands are affected in only 8% of the cases.[1] Patients with intraoral pleomorphic adenomas in the upper lip rarely present to the otolaryngologists since such lesions are simply excised surgically before causing any cosmetic deformities.[2-4] We present a rare case of giant pleomorphic adenoma in the upper lip region, with erosion of the maxillary bone and distortion of the facial appearance.

CASE REPORT

A 20-year-old man presented with a painless mass on his upper lip, which had gradually increased in size over a period of 2 years. By the time the patient had been referred to our department, the huge mass on his upper lip had distorted his appearance. Bimanual palpation revealed a nontender, mobile 4 cm × 3 cm submucosal mass superior to the vermillion border of the upper lip and exerting pressure on the patient's cheek and the anterior wall of the right maxillary sinus [Figure 1]. The overlying mucosa was normal in appearance, and there were no palpable lymph nodes. Computed tomography demonstrated a 30 mm × 28 mm enhancing mass in the upper lip region with no invasion to the surrounding tissues. Erosion of the maxillary bone posterior to this lesion was noted [Figure 2]. After patient's written consent was taken aspiration biopsy was performed. Fine-needle aspiration biopsy of the mass was consistent with the diagnosis of a benign mixed salivary gland tumor. Under general anesthesia, the lesion was excised completely with a wedge of mucosa overlying the tumoral mass. Sufficient abdominal fat was harvested through a 15-mm inferior umbilical incision and inserted into excision site as a volume filler. There was no significant bleeding, and after suturation, the mucosal flaps were well approximated. Healing was uneventful, and no recurrence or facial asymmetry was observed after 1 year of follow-up. Histological examination revealed a well-circumscribed submucosal lobular tumor consisting of ductal tubules lined by cuboidal cells, with adjacent areas of

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chondroid and myxoid stroma and proliferation of clusters of epithelial cells. No cytologic atypia or mitotic figures were observed [Figure 3]. These pathological findings confirmed the diagnosis of a minor salivary gland pleomorphic adenoma in the upper lip.

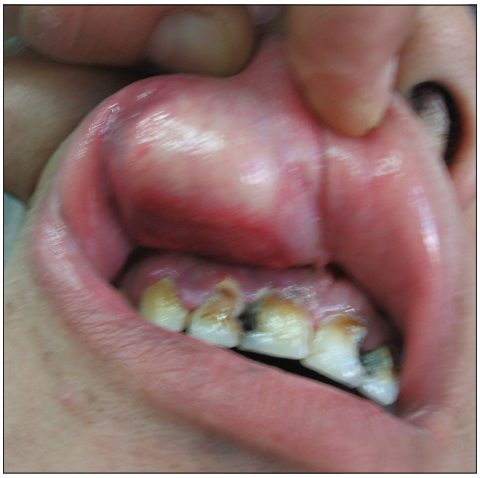

Figure 1: The appearance of the lesion in the upper lip

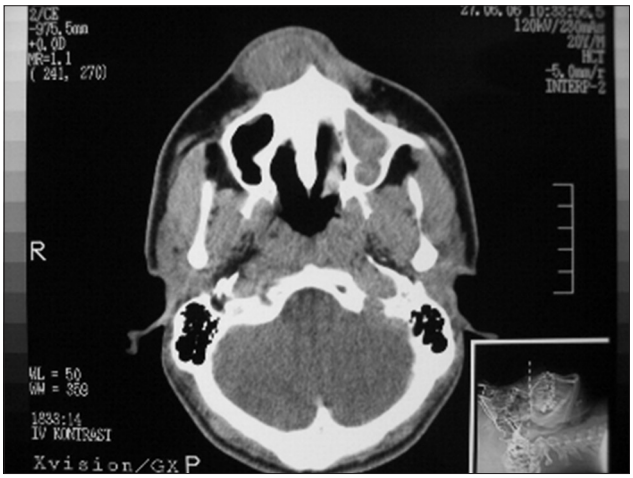

Figure 2: Computed tomography demonstrating a 30 mm × 28 mm enhancing mass in the upper lip region with erosion of the maxillary bone

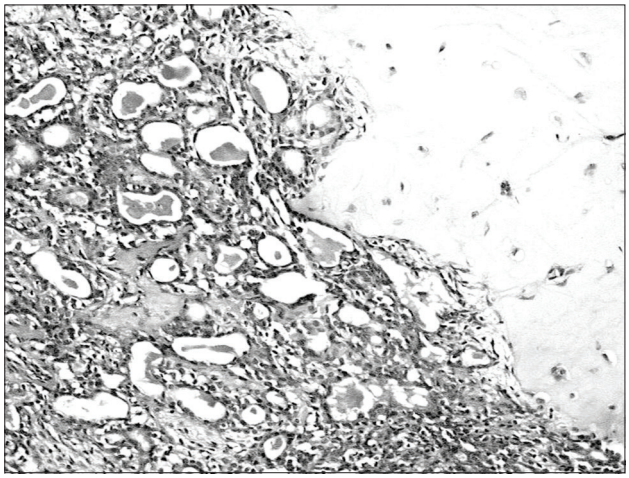

Figure 3: Microphotograph showing characteristic ductal tubules which are lined by cuboidal cells and contain eosinophilic material

DISCUSSION

Salivary gland tumors represent about 3% of all head-and-neck neoplasms. Approximately 80% are located in the parotid gland, 10% in the submandibular gland, and the remainder being distributed between the sublingual gland and the countless minor salivary glands. The last group comprises the innumerable tiny submucosal serous and mucinous glands to be found in the oral cavity, nose, sinuses, postnasal space, oropharynx, larynx, and trachea.[5] Pleomorphic adenoma (benign mixed tumor) is the most common neoplasm of salivary gland origin. They constitute 60% of the benign tumors from all salivary gland sites: 61% of major gland tumors and 54% of minor gland tumors.[6] Minor salivary gland pleomorphic adenomas present over a wide age range, but presentation in the fourth decade is the most common. In the previous reports, the male-to-female ratio for this tumor ranged from 1:1.1[7] to 1:2.5.[8] The palate is the most common site for intraoral pleomorphic adenomas, followed by buccal region and upper lip, respectively.[3,4] There is a propensity for benign tumor to occur in the upper lip, besides malignant lesions are often seen in lower lip.[9] Mixed tumors typically present as slow-growing, asymptomatic, nodular masses stretching the overlying skin or mucosa. Intraoral swelling is the most frequent of the signs and symptoms of minor salivary gland tumors. Ulceration, pain, and change in sensation are uncommon features. They vary in consistency from soft and fluctuant to firm and rubbery, depending on the presence of cystic and mucoid degeneration or the formation of chondroid and osteoid tissues.[10] The microscopic diagnosis of the lesions is generally easy. Tubuloalveolar and gland-like structures with two or more cuboidal cell lines, islands of cuboidal or polygonal cells in a fibroadipose, chondroid, hyaline, or mucinous hypocellular stroma that is positively stained for periodic acid-Schiff and alcian blue are indicative features. Mixed tumors that originate from the glands in the oral mucosa frequently have only a poorly developed or no capsule, although these tumors usually readily separate from surrounding tissue during surgery.[6] Distinguishing salivary gland mixed tumor (pleomorphic adenoma) from dermal mixed tumor (chondroid syringoma) may be difficult if not impossible. This dilemma occurs most often in specimens from the upper lip. The presence of salivary gland tissue adjacent to the tumor and the clinical presentation of a mucosal rather than dermal protuberance lend some support to a salivary origin.[6] However, the clinical presentation and the histopathological features of the present case were consistent with the diagnosis of pleomorphic adenoma in the upper lip.

In summary, pleomorphic adenoma of the upper lip is an uncommon intraoral tumor which should be taken into consideration in the differential diagnosis of any slowly growing mass in the lip region. The correct diagnosis is established by a thorough histological examination of the tumor. Local surgical excision with surrounding soft tissue is the treatment of choice, and the histological specimen must be examined closely to confirm complete excision and to ascertain that there is no evidence of malignant change.

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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