

Pleomorphic Adenoma of the Upper Lip: A Case Report and Review of Literature

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ABSTRACT

Pleomorphic adenoma, 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 pleomorphic adenoma of the minor salivary glands are the palates followed by lips and cheeks. Here we report a case of this tumor presenting in minor salivary glands of upper lip in a 16 year old girl. The patient presented with a painless, mobile, circumscribed nodule, about 1.5 cm in diameter, slowly growing in size. The nodule was subjected to Fine Needle Aspiration Cytology (FNAC) and was found to be pleomorphic adenoma of minor salivary gland origin in upper lip. Complete excision was performed and the diagnosis was confirmed by histopathological examination. The aim of this article is to highlight the occurrence of pleomorphic adenoma as a differential diagnosis for the swellings in the upper lip.

Keywords: Pleomorphic adenoma, minor salivary gland tumors, upper lip

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INTRODUCTION

Pleomorphic adenoma (PA) is the most common benign salivary gland neoplasm. It is usually found in the parotid gland but may occur in the submandibular, sublingual and minor salivary glands¹. Tumors of the minor salivary glands are uncommon and account for 10-25% of all salivary gland tumors. Pleomorphic adenoma is

the most common neoplasm of the minor salivary glands². The palate is considered the most common intraoral site (42.8-68.8%), followed by the 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 mucosa³. It mainly affects women in their fourth to sixth decades. The

patients usually present with a painless and slow-growing mass⁴]. Pleomorphic adenoma arising from minor salivary glands of the lips tends to occur at an earlier age than it does at other sites⁵]. 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 are responsible for the production of abundant, extracellular matrix with chondroid, collagenous, mucoid and osseous stroma⁶]. We present a rare case of

Pleomorphic adenoma in the upper lip of 16 year old girl. A brief review of the relevant literature is also presented.

CASE REPORT

A 16 year old female presented in Ear Nose and Throat Out Patient Department of this hospital with the complaint of painless, mobile, nodular swelling in upper lip. The nodule slowly increased in size during the past three months. On examination, the mass was non tender, circumscribed, mobile, sessile and rubbery in consistency and 1.5 cm in diameter (Figure 1).

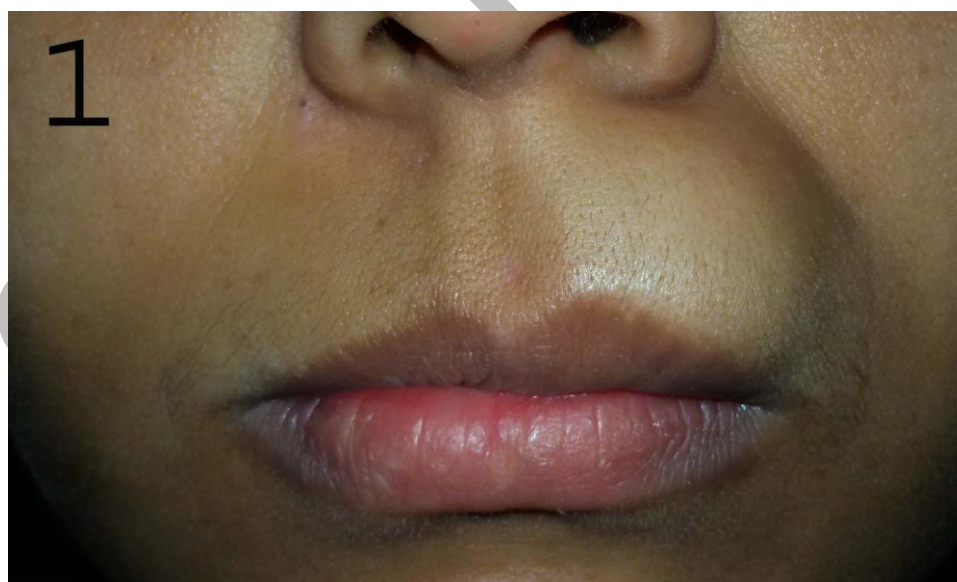


Figure 1- Clinical photograph of a nodular, circumscribed swelling of upper lip measuring 1.5cm.

Skin over the tumor was not fixed. The lip mucosa was unremarkable. Head and neck abnormalities were not noted

on clinical examination. There was no preceding history of trauma and her past medical history was

unremarkable. Fine Needle Aspiration Cytology revealed a biphasic pattern comprising epithelial component and

fibromyxoid stroma, suggesting a diagnosis of pleomorphic adenoma (Figure 2a).

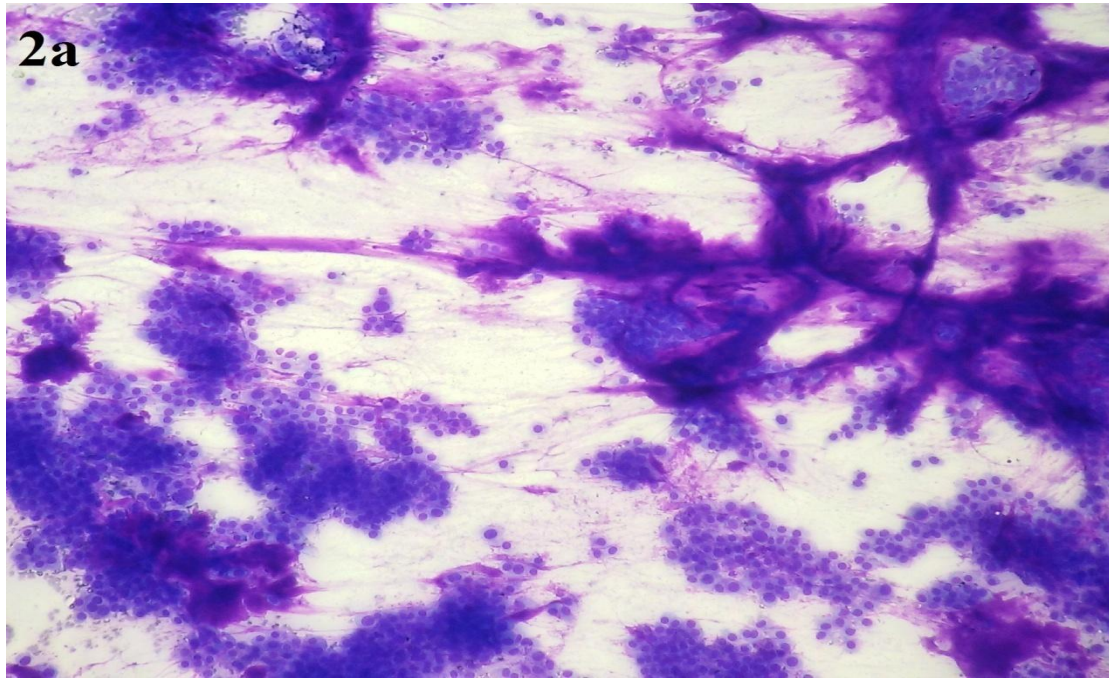


Figure 2a- FNA smears reveal sheets of epithelial cells and abundant fibromyxoid stroma (MGG; x100).

The tumor was completely excised; the mass was fully encapsulated without any subcutaneous or muscle attachment. Grossly, a grey white tissue piece of firm consistency was received, measuring 1.5x1x0.5 cm. The cut surface of the lesion was solid and grey white to tan in colour with occasional minute cystic areas.

Histopathological analysis of the surgical specimen revealed a circumscribed tumor composed of biphasic population of cells comprising of nests and islands of epithelial cells which appeared to be melting into the stroma. Foci of cartilage and mesenchymal proliferation were present (Figure 2b).

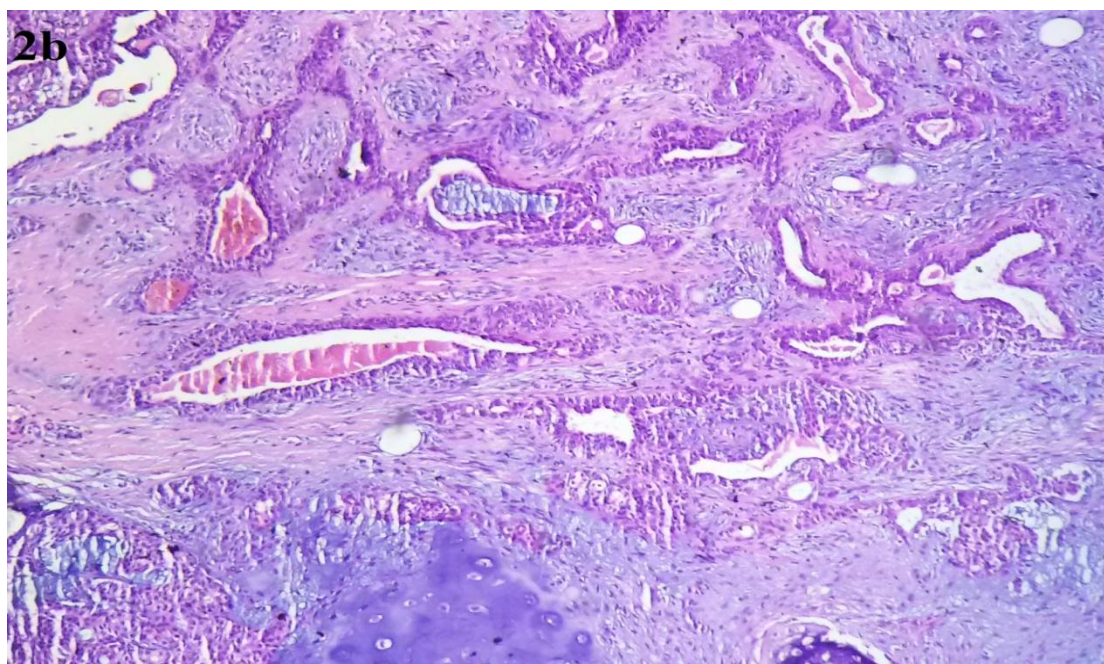


Figure 2b- Microsections reveal nests and islands of epithelial cells appearing to be melting into the stroma. Foci of cartilage also present (H and E; x100).

There was no evidence of malignancy. The diagnosis given was pleomorphic adenoma of upper lip.

DISCUSSION

Salivary gland tumors mainly occur in adults, being rarely reported in children. Less than 5% of all pediatric head and neck tumors arise from salivary glands. Minor salivary gland tumors are rare in children and are responsible for only 5% of all salivary gland tumors⁶]. However, Lotufo et al⁶] and Jorge et al⁷] reported PA of the upper lip in three patients under the age of 18. According to Bradley et al⁸], pediatric salivary gland epithelial neoplasms usually occur between 7 and 16 years of age, with an average age of 10.5 years. The peak incidence

of PA arising from minor salivary glands of the lips is in the third and fourth decades of life, with an average of 33.2 years. In addition, there is a propensity for malignant tumors to occur on the lower lip, whereas benign lesions predominate on the upper lip. This finding may be related to the differences in embryonic development between the lower and upper lips^{9, 10}]. The definite etiology is not known. However, cytogenetic and molecular studies have shown an epithelial origin for the tumor with chromosomal abnormality in 8q12 and 12q25⁴]. Pleomorphic adenoma of the upper lip

exceeds that of the lower lip by the ratio of 6:1⁵]. Eveson and Cawson documented 75% of upper lip tumors as benign¹¹]. Minor salivary gland tumors present as soft or firm masses, with most having a nodular, exophytic component.

Ulceration of the nodular mass may occur, but the presence of ulcer provides no clue to the invasiveness of the tumor. Those that are soft on palpation usually have large cystic cavities and an abundance of mucin. The more solid tumors, especially pleomorphic adenoma with bone and cartilage formation, are firm on palpation⁵].

Minor salivary gland tumors present as soft or firm masses, with most having a nodular, exophytic component. Pleomorphic adenomas are usually painless, slow growing tumors; however rare cases exhibiting rapid growth have been reported, especially in the palate. The chances of recurrence are higher when pleomorphic adenoma occurs before 30 years of age⁶].

Differential diagnosis of a clinically benign mass of the upper lip can be classified into three groups: (1) salivary gland tumors such as

Pleomorphic adenoma, canalicular and mucoepidermoid carcinomas; (2) mesenchymal tumors such as lipoma, leiomyoma, nerve tumors (neurofibroma, neurilemmoma/schwannoma), benign fibrous histiocytoma, oral focal mucinosis and granular cell tumor; and (3) infections such as tuberculosis, syphilitic gumma (if the lesion presents as an ulcerated mass) and deep fungal infections

(histoplasmosis, cryptococcosis, blastomycosis, coccidioidomycosis)¹²].

The surgical treatment for pleomorphic adenoma is complete wide excision with a good safety margin.

Pleomorphic adenoma appears to be encapsulated, but this capsule is often infiltrated by lateral extension of tumor. Even though pleomorphic adenoma is benign, it has high rate of implantability. Any rupture of the capsule or incomplete excision or tumor spillage during excision will leave residual tumor cells behind, resulting in recurrence¹³]. Therefore a long term follow up is necessary to look for recurrence and/ or malignant transformation.

CONCLUSION

Pleomorphic adenoma in minor salivary gland of upper lip is very rare. Fine Needle Aspiration Cytology is a valuable diagnostic adjuvant in preoperative evaluation of salivary gland lesions. It provides preliminary diagnosis and preoperative assessment on which management decisions can be based.

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