Case Report

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PLEOMORPHIC ADENOMA OF UPPER LIP: CASE REPORT

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ABSTRACT

Pleomorphic adenoma is the most frequently encountered salivary gland tumor, with 90% of cases occurring in major salivary gland and 10% in minor salivary glands. There is an annual incidence rate of approximately 2.4 to 3.05 per 100,000 people worldwide. The most common site of occurrence in minor salivary glands is upper lip and palate. Pleomorphic adenomas of the minor salivary glands typically present as painless, submucosal swellings. Adequate surgical excision is the treatment of choice to avoid any recurrence. We report a case of swelling of upper lip in a 45 years old female patient, which histopathologically was diagnosed as pleomorphic adenoma, with emphasis on the differential diagnosis.

KEYWORDS: Salivary gland; swelling, tumor

INTRODUCTION

The pleomorphic adenoma is the commonest of the salivary gland tumors, accounting for 50-70% 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% cases). The lesion appears as a painless, slow growing, firm mass. The tumor occurs at any age but most commonly between the ages of 30 and 50 with female predilection.[1] We report a rare case of swelling occurring in upper lip of a middle aged female patient which was mimicking a fibrosed mucocele, but histopathologically was diagnosed as pleomorphic adenoma.

CASE REPORT

A 45 years old female patient reported to us with a chief complaint of swelling of the upper lip (Fig 1). No significant associated history was present, except history of lip biting since many years. The swelling was gradual in onset with slowly increasing size with no pain or discharge. There was discomfort in chewing and speaking. General physical examination and medical examination of the patient did not reveal any abnormality. Extraoral examination revealed swelling of the left side of upper lip. Intraoral inspection showed well defined solitary, lobulated swelling measuring 1x2cm in diameter in the upper lip, extending superior-inferiorly from the depth of the vestibule to the vermillion border of the upper lip and medio-laterally 0.5cm from the labial frenum to the left corner of the mouth (Fig 2a). Overlying mucosa was of same color as that of adjacent mucosa, with smooth surface and no ulceration. On palpation, it was non-tender, freely movable and firm in consistency (Fig 2b). Blood examination reports were within normal limits. Based on the history and clinical examination of patient a provisional diagnosis of mucocele undergoing fibrosis was given. As the lesion was well circumscribed, an excisional biopsy under local anesthesia was considered. An elliptical incision was made around tumor including 2mm of fresh margins. The lesion was excised in *toto* and sutures were placed maintaining the contour of the lip, once homeostasis was achieved (Fig. 3). Histopathology revealed an encapsulated lesion composed of epitheloid type of myoepithelial cells arranged in cords and network like pattern in a connective tissue stroma which was hyalinized at few places and myxoid at others. Epithelial cells arranged in cords, strands and islands forming ductal and cystic structures were seen. These ductal structures were bilayered at places and at few places lined by single layer of cells. The cystic structures were filled with eosinophilic material. Few areas showed squamous metaplasia with keratin pearl formation (Fig 4). On the basis of histopathological

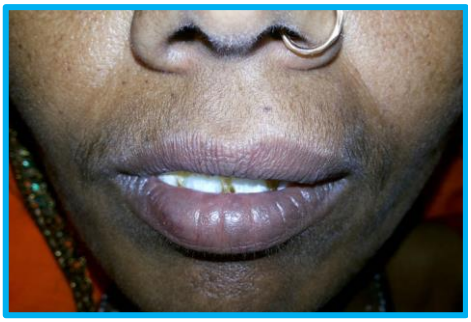

Fig. 1: Extraoral examination showing swelling of left side of upper lip

Fig. 2: Intraoral examination showing extent of the lesion

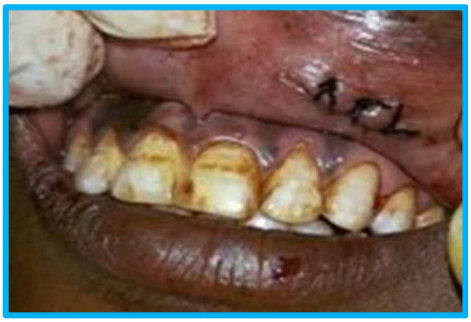

Fig. 1: Post operative picture showing complete surgical excision with sutures in place

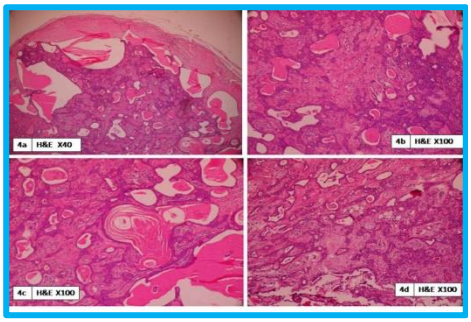

Fig. 4: Photomicrographs showing a) well encapsulated lesion showing epithelial cells in cords and island with ductal and cystic structures (H&E stain X40); b) multiple cords of epitheloid type of myoepithelial cells around ductal structures which are filled with eosinophilic material (H&E stain X100); c) squamous metaplasia with keratin pearl formation (H&E stain X100); d) myxoid and hyalinized areas (H & E stain X100)

examination, final diagnosis of pleomorphic adenoma was made.

DISCUSSION

Salivary gland tumors are rare, constituting 2–6.5% of all head and neck tumors. Tumors of the minor salivary gland account for 22% of all salivary gland neoplasms. Pleomorphic adenoma is the most common tumor of the salivary gland making up 70% of the parotid tumors, 55% submandibular gland tumors and approximately 10% of the minor salivary gland tumors.[1] The most common site for pleomorphic adenoma of the minor salivary glands is the palate, which is followed by the upper lip. No specific associated etiology is known. But, the molecular and cytogenetic studies done for the tumor have shown an epithelial origin with chromosomal defect in 12q25 and 8q12.[2] Pleomorphic adenoma of the minor salivary glands is not completely capsulated and is characterized microscopically by cellular pleomorphism. But, in the present case, the tumor was completely encapsulated. Epithelial and myoepithelial elements blend most commonly with tissue of myxoid, chondroid or mucoid appearances.[1] Synonyms include Mixed Tumor, Enclavoma, Endothelioma, Enchondroma, and Branchioma. In view of atypical histological patterns of the lesion, the tumor was named Pleomorphic aden-oma by Willis.[1] Pleomorphic adenoma arising from lips tends to occur at an earlier age than does at other sites. Bernier found that the peak incidence of pleomorphic adenoma of the lips was in third to fourth decade of life, with an average age of 33.2 years. Pleomorphic adenomas present as slow-growing, painless mass, forming smooth, firm and mobile lumps.[3] This similar presentation was noted in the present case also. The differential diagnosis of well defined submucosal swelling in the upper lip should be arrived based on the anatomic structures present in that location. The lesions included in the differential diagnosis were Mucocele, Irritational fibroma, Schwannoma, Neurofibroma and Canalicular adenoma.[4,5] The most common lesion on lip is mucocele which is associated with history of trauma to lip. But the commonest site is lower lip where it clinically appears as soft, fluctuant, bluish colored swelling. Although history of trauma to upper lip was present, but

mucocele rarely occurs on upper lip. But since the duration of trauma to lip has been present since many years so there was a possibility of this lesion to be a mucocele undergoing fibrosis. So this was considered in the differential diagnosis. Irritational fibromas can also be included in the differential diagnosis. But this lesion mostly occurs on the gingiva, buccal mucosa and tongue. Though Schwannoma and neurofibromas are quite common in oral cavity presenting as submucosal mass, but both have younger age predilection and most common site is tongue. In upper lip, the most common salivary gland tumor is canalicular adenoma, typically presenting in older age females as painless, slow growing, and freely movable, firm masses.[4,5] But this lesion rarely occurs in the oral cavity. Histologically, Pleomorphic adenomas are characterized by a remarkable degree of morphologic diversity between individual tumors and often within a single tumor mass. The most important histologic components of the tumor are the epithelial and myoepithelial cells with the stromal elements. HUMARA assay have shown a common origin for stromal and epithelial cells of pleomorphic adenoma.[6] The epithelial component of pleomorphic adenomas shows a wide variety of cell types, including squamous, basaloid or cuboidal. The myoepithelial cells can be either clear, plasmacytoid or spindle. The epithelium can form cords, sheets or ductal structures. The myoepithelial cells may be similar to the duct lining cells or have eosinophilic to clear cytoplasm with small, flattened and hyperchromatic nuclei. The ducts can include eosinophilic material and form microcysts. More solid areas of epithelial cells lacking ductal lumini may be seen admixed with myoepithelial cells. The mesenchymal component which is a product of modified myoepithelial cells can be myxoid or hyalinized, or cartilaginous. Squamous metaplasia is also noted. Cells within the myxoid areas are myoepithelial in origin. Bone may form by stromal osseous metaplasia.[7] Though immunohistochemistry is rarely of value in differential diagnosis, a number of immunocytochemical studies have been done to provide insights into the histogenesis of this unique tumor. Modified myoepithelial cells are irregularly positive for pan-cytokeratin and cytokeratins 13, 14 and 16.[7] They are also positive for calponin, vimentin, S-100, α-smooth muscle actin and glial fibrillary acidic protein. The non lacunar cells of chondroid areas are positive for both vimentin and pan-cytokeratin, whereas lacunar cells are positive only for vimentin.[7] Minor salivary gland tumors are treated by a complete surgical excision. Prognosis is excellent. Recurrence may be due to incomplete excision.[8]

CONCLUSION

Pleomorphic adenoma of the upper lip is an unusual neoplasm. A complete surgical excision is the treatment of choice. Regular follow up is necessary to check for recurrence and malignant transformation.

CONFLICT OF INTEREST & SOURCE OF FUNDING

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