

PLEOMORPHIC ADENOMA OF MINOR SALIVARY GLAND OF CHEEK: A RARE CASE REPORT AND REVIEW OF LITERATURE

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ABSTRACT

Pleomorphic adenoma is the most common benign salivary gland neoplasm. The involvement of minor salivary glands of cheek is extremely uncommon and accounts only for 3-6%. We are presenting here such a rare case of pleomorphic adenoma of minor salivary gland of cheek in an adult female who presented to ENT OPD with complaint of right cheek swelling from last 2 years. On clinical examination swelling was firm, non tender, freely mobile with normal overlying skin and inner buccal mucosa. As a part of evaluation protocol, USG cheek and FNAC was done. The swelling was excised and sent for histopathological examination which came out as pleomorphic adenoma arising from minor salivary gland of cheek. The patient was followed for 1 year post operatively without any recurrence.

KEYWORDS: Buccal mucosa, minor salivary gland, pleomorphic adenoma.

INTRODUCTION

Salivary gland tumors are uncommon and constitute 2–6.5% of all the head and neck neoplasm.^[1] Pleomorphic adenoma is the most common benign neoplasm of the salivary glands. The palate is considered as the most common intraoral site, followed by the upper lip and cheek.^[2] Pleomorphic adenoma consists of cells with epithelial and mesenchymal differentiation (mixed tumor). Now it is widely accepted that both epithelial and mesenchymal (myxoid, hyaline, chondroid, osseous) elements often arise from same cell clone either myoepithelial or ductal reserve cell.^[3] Pleomorphic adenoma usually presents as a mobile slowly growing, painless firm swelling without any ulceration of the overlying mucosa.^[4] This tumor usually present in the fourth to sixth decades with a slight female predominance. In about 5–10% cases, minor salivary gland pleomorphic adenomas is seen in patients less than 20 years of age. Cases have been reported in the literature in patients as young as 3 months up to 18 years.^[5,6,7] The mucosa of the cheek is an uncommon site for intraoral pleomorphic adenoma and this case is being reported her for its rarity.

CASE PRESENTATION

A 30 year old female patient came to our department with chief complaint of swelling and pain in right cheek from last 1 year. There was no history of fever, dysphagia or dental trauma. Intraoral examination including digital palpation revealed a 2.5 cm × 2 cm well circumscribed, freely mobile, firm and non tender

solitary mass over the right cheek with normal overlying mucosa (Figure 1). There was no previous history of any similar swelling in the same region. As a part of evaluation protocol USG cheek and FNAC was done and a provisional diagnosis of pleomorphic adenoma of minor salivary gland was made. The tumor was excised via intraoral approach under local anesthesia. The mass was found to lie between the buccal mucosa and buccinators muscle. After meticulous dissection, the lesion was freed from the surrounding tissue and was removed along and sent for histopathological examination. The mass had a thin capsule and cut surface shows grayish white appearance.

Histological examination showed chondromyxoid areas with interspersed tubules and sheets of cells consistent with pleomorphic adenoma (Figure 2). Postoperative period was uneventful. Patient is under regular follow up without any recurrence.

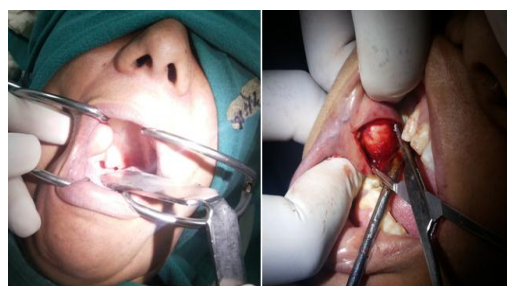


Figure 1: Showing pre and intraoperative pics of swelling

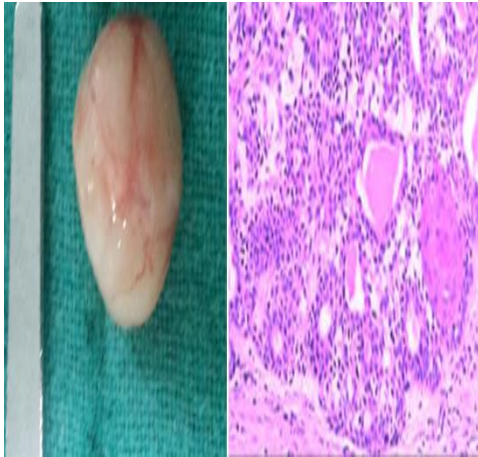


Figure 2: Showing gross and microscopic picture of the tumor

DISCUSSION

Pleomorphic adenoma is the most common minor salivary gland tumor accounting for about 33–70% of all tumors and 70.6–100% of benign tumors. The palatal glands are the most commonly affected with pleomorphic adenoma with a frequency ranging from 43% to 70% 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 mucosa.^[5,6,7,] The pleomorphic adenomas in the buccal mucosa are known to arise from the submucosal buccal minor salivary glands.^[8] This tumor usually presents as a unilateral, painless, slow growing mass in the parotid gland. Few cases are also reported in the literature with ulceration, pain and bleeding.^[6,7] The pleomorphic adenomas in youngsters have similar biological characteristics as in adults but with low recurrence rates after surgical excision.^[6] In our case, it was presented as a swelling associated with normal overlying mucosa. The differential diagnosis of minor salivary gland pleomorphic adenoma of the cheek includes buccal space abscess, hemangioma, dermoid cyst, lipoma, neurofibroma, rhabdomyosarcoma, mucoepidermoid carcinoma, adenoid cystic carcinoma, carcinoma ex pleomorphic adenoma. Histologically, these tumors will have epithelial cells arranged in cord or duct like cell patterns, along with epidermoid metaplasia. The intercellular matrix shows fibrous, hyaline, myxoid, cartilaginous, and osseous areas. Histological variants of pleomorphic adenoma include pleomorphic adenoma with lipomatous change, myxolipomatous pleomorphic adenoma, pleomorphic adenoma with squamous differentiation, and benign metastasizing mixed tumor.^[9] Treatment of pleomorphic adenoma includes excision with a margin, and use of adjuvant radiotherapy. Many pleomorphic adenomas of minor salivary glands will have a capsule that is either thin or incomplete.^[6,9] Though these benign tumors are encapsulated, they require resection with an adequate margin of normal surrounding tissue because these tumors are known to have microscopic pseudopod like extensions into the surrounding tissue due to dehiscences in the false

capsule. Hence, incisional biopsy is avoided in these tumors to avoid spillage of the tumor cells.

The recurrence of this tumor is a well known factor. Lou *et al.* reported a recurrence of 7% in 1342 patients with benign parotid neoplasm and 6% in benign minor salivary gland tumors. Recurrences are reported even at 18 years after initial treatment. Inadequate initial surgical procedure was reported to be the main cause of failure.^[7,8] In recent years, immunohistochemical markers are playing a major role in distinguishing pleomorphic adenomas from malignant salivary gland tumors. Human epidermal growth factor receptor-2/neu expression, androgen receptor expression, overexpression of P53, and expression of Ki-67 can be used in evaluating malignant salivary gland tumors, but these markers may also be expressed in benign pleomorphic adenomas. Hence, in the absence of compelling histologic features of malignancy, expression of these immunohistochemical markers should not be interpreted as evidence of carcinomatous transformation in a pleomorphic adenoma.^[10]

CONCLUSION

Pleomorphic adenoma of the cheek is a rare neoplasm and diagnosis requires a high index of suspicion. Complete wide surgical excision is the treatment of choice. Recurrence as well as malignant transformation even after many years of surgical excision should be kept in mind and therefore a long term follow up of these cases is advisable.

CONFLICT OF INTEREST

No financial or conflict of interest

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