PLEOMORPHIC ADENOMA OF MINOR SALIVARY GLANDS IN CHEEK: A CASE SERIES

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ABSTRACT
The pleomorphic adenoma, also known as the benign mixed tumour comprises the majority of both major and minor salivary gland tumours, accounting for 3-10% of neoplasm of the head and neck region. The palate is the predominant intraoral site and the mucosa of the cheek is an uncommon site for these tumours. Review of literature, clinical features, pathology and treatment of these tumours in 5 such cases are discussed and reported in the present case series.

KEYWORDS: Pleomorphic adenoma, cheek, minor salivary glands, benign salivary gland tumours.

INTRODUCTION
Pleomorphic adenomas (PAs) are benign salivary gland tumours that represent about 3-10% of the neoplasm of the head and neck region.¹,² PA is the most common benign neoplasm of the salivary glands. About 90% of these tumors occur in the parotid gland and 10% in the minor salivary glands. Among the minor salivary gland tumours, palate is considered as the most common intraoral site (42.8-68.8%), followed by the upper lip (10.1%) and cheek (5.5%).³ Other rare sites include the throat (2.5%), retromolar region (0.7%), floor of the mouth and the alveolar mucosa.⁴ They are more common in adult females from the 3rd to 5th decades.⁵

PA is of glandular origin, usually presenting as a slowly growing, painless, firm swelling that does not cause ulceration of the overlying mucosa.⁶ Histologically, PA consists of cells with epithelial and mesenchymal differentiation. The treatment of choice for PA is surgical removal with safety margins, to prevent the recurrence. Recurrence rate of 5-30% has been found for PA, so a periodic follow-up is must, due to the important relapse potential and aggressiveness of these lesions.⁷

As the mucosa of the cheek is a relatively rare site of occurrence for intraoral pleomorphic adenoma, the authors present a series of 5 such cases, reported in the department of Otorhinolaryngology at B.P.S Government medical college for women, Khanpur Kalan, a rural tertiary care centre.

CASE SERIES
All 5 patients reported, visited in the ENT outpatient department of our institute from where they were sent to department of radiology for ultrasound examination (USG) of cheek and the department of pathology for fine needle aspiration cytology (FNAC). The clinical, radiological and histopathological features of the patients are given in (Table 1).

Clinical presentation
Out of the 5 patients, 4 were females and 1 was male. Age ranged from 18 to 48 years. All patients presented with the complaints of painless swelling over the cheek for last 6 months to 18 months. None of the cases had associated ulceration or any discharge. There was no history of trauma, fevers, disturbance of salivation, or oral surgeries. General examination of all patients did not reveal any significant findings. No history of chronic alcoholism or tobacco was given by any patient. There was no previous history of any similar swelling in the same region. On examination, in all the cases, there was a firm, smooth, non-tender, well circumscribed lesion in the cheek. Overlying skin and mucosa was healthy. No significant lymphadenopathy was found in the neck region. The laboratory tests were unremarkable.

Cytological features
Fine needle aspiration cytology of the lesion was performed and the smears were stained with Giemsa stain. Smears revealed the presence of bimodal pattern of epithelial cells and spindle cells in a myxoid stroma. The epithelial cells were of uniform size with round to oval nuclei, moderate amount of cytoplasm and well defined cell boundaries. A diagnosis of pleomorphic adenoma was made. All patients had surgery for removal of mass, after meticulous dissection, the lesion was freed from the surrounding tissue and was sent for histopathological examination. The mass had a thin capsule and cut surface shows whitish appearance.
Postoperative period was uneventful. All Patients are under regular follow up and are without any recurrence yet.

**Histopathological features**
Histopathological examination revealed a well encapsulated tumor outside which small amount of normal salivary gland tissue was seen. Tumor tissue consisted of gland like structures and sheets of epithelial cells with myxoid and chondroid areas. No mitotic figures were found. These features were consistent with diagnosis of a pleomorphic adenoma.
DISCUSSION

Pleomorphic adenoma occurs more in females with the ratio range from 1.2:1 to 1.9:1 in previous studies\cite{7,10} and is most common from the third to fifth decade. Study done by Moshy et al. showed slight predilection for older patients.\cite{11} On the other hand, study done by Waldrom et al. revealed that tumor was more common in younger age group.\cite{12} The most common mean age range is 43-46 years.\cite{4} In the present study, occurrence is more in females with Female to Male ratio of 1.67:1. The mean age of incidence in the present study is 35 years with the range of 18-45 years.

The PA of minor salivary glands clinically present as painless, slow growing sub mucosal masses. The covering mucosa is seldom affected unless it is secondarily traumatized. The majority of intraoral mixed tumors are less than 3.0 cm in diameter. They are usually solitary and well-circumscribed. The findings of the case

<table>
<thead>
<tr>
<th>S.No.</th>
<th>AGE</th>
<th>SEX</th>
<th>SITE</th>
<th>SIZE (cm)</th>
<th>PAIN</th>
<th>CONSISTENCY</th>
<th>HISTOPATHOLOGY</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>18</td>
<td>Female</td>
<td>L cheek</td>
<td>3*2.5</td>
<td>-</td>
<td>Firm</td>
<td>Fibromyxoid Stroma, glandular epithelium arranged in sheet pattern</td>
</tr>
<tr>
<td>2</td>
<td>25</td>
<td>Female</td>
<td>L cheek</td>
<td>2.5*2.5</td>
<td>-</td>
<td>Firm</td>
<td>Fibromyxoid Stroma, glandular epithelium arranged in sheet pattern</td>
</tr>
<tr>
<td>3</td>
<td>45</td>
<td>Male</td>
<td>L cheek</td>
<td>3*3</td>
<td>-</td>
<td>Firm</td>
<td>Chondromyxoid Stroma, glandular epithelium arranged in sheet pattern</td>
</tr>
<tr>
<td>4</td>
<td>43</td>
<td>Female</td>
<td>R cheek</td>
<td>2.5*3</td>
<td>-</td>
<td>Firm</td>
<td>Fibromyxoid Stroma, glandular epithelium arranged in sheet pattern</td>
</tr>
<tr>
<td>5</td>
<td>44</td>
<td>Male</td>
<td>L cheek</td>
<td>3*4</td>
<td>+/-</td>
<td>Firm</td>
<td>Fibromyxoid Stroma, glandular epithelium arranged in sheet pattern</td>
</tr>
</tbody>
</table>
presented here is in agreement with those of other investigators.[5,6,8]

In the present study, Histological examination showed cells arranged in sheets and tubules intermixed with fibromyxoid and chondromyxoid stroma consistent with the intercellular matrix shows fibrous, hyaline, myxoid, cartilaginous and osseous areas reported in previous study.[3]

The differential diagnosis of PA cheek includes buccal abscess, dermoid cyst, sebaceous cyst, neurofibroma, lipoma, mucoepidermoid carcinoma and polymorphous low grade adenocarcinoma.[10] The possibility of buccal space abscess was ruled out due to absence of sign of inflammation. The solid nature of lesion coupled with lack of tissue representing the three germ layers rule out the possibility of mature dermoid cyst. Absence of punctum and freely movable nature of the mass differentiate PA from sebaceous cyst. As on histological picture both epithelial and myoepithelial cells were seen, which rules out mucoepidermoid carcinoma. The negative slip test clinically and absence of lipomatous component histologically rules out lipoma. The absence of perineural invasion and mitotic figures obscure the chances of polymorphic low grade adenocarcinoma.[9] The ideal treatment of choice for PA is wide local excision with good safety margins and follows-up for at least 3–4 years.[8] In the present series, wide local excision was done in all cases and cases are in regular follow up.

CONCLUSION
Pleomorphic adenoma of the cheek is a rare neoplasm and therefore its diagnosis requires a high index of suspicion. Pleomorphic adenoma should always be considered in the differential diagnosis of cheek masses. Complete wide surgical excision is the treatment of choice. Since the majority of minor salivary gland tumors are reported to be malignant, careful history, patient evaluation, histopathological examination is advised.

REFERENCES