Intracapsular Carcinoma ex Pleomorphic Adenoma in the Buccal Space: Management Dilemma

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Abstract
Objective: Rare case of intracapsular carcinoma ex pleomorphic adenoma in the buccal space with focus on its appropriate management.
Method: Case report with review of current literature on buccal space lesions and intracapsular carcinoma ex pleomorphic adenoma.
Result: Although salivary gland tumors are common, intracapsular carcinoma ex pleomorphic adenoma is a rare entity especially within the buccal space with no similar case report in the current literature.
Conclusion: Our case is unusual due to its unusual presentation in the buccal space and pathological features. This case report also highlights issues regarding its appropriate management.
Keywords: Pleomorphic adenoma, salivary gland neoplasm, facial neoplasm.

CASE REPORT

A 50 years old male presented with a right sided cheek swelling of 3 months duration. It was nonprogressive and painless. There was no history of previous surgical intervention. On palpation, the swelling was smooth, nodular, firm, free from the surrounding structures and close to the oral mucosa. Thus, clinically it appeared like a benign tumor in the buccal space. Magnetic resonance imaging was also corroborative revealing a 3.2 cm, moderately vascular, well-encapsulated tumor within the buccal space without any infiltration into the surrounding structures (Fig. 1).

Fine needle aspiration however, suggested a poorly differentiated adenocarcinoma. Hence a wide excision of the tumor was planned. Since it was palpable close to the oral mucosa, an intraoral approach was adopted. Frozen section suggested a pleomorphic adenoma. However, the final histopathology revealed a carcinoma ex pleomorphic...
adenoma. Since the clinical findings did not corroborate with the histopathology, the slides were reviewed by a senior pathologist. It was found that the histopathology was indeed pleomorphic adenoma with focal intracapsular carcinomatous changes (Figs 2 and 3). Immunohistochemistry stained positively for cytokeratin (CK) and epithelial membrane antigen (EMA). In view of the histopathology suggestive of carcinomatous changes, patient was subjected to adjuvant radiotherapy after a literature search did not suggest any separate management guidelines for intracapsular carcinoma ex pleomorphic adenoma.

The patient received radiotherapy in 30 fractions with a dose of 2 grays/fraction. Patient is loco regionally controlled at one year of follow-up.

DISCUSSION

Buccal space is confined to the cheek lateral to the buccinator. It was first described in 1935 by Coller and Yglesias. It is an anatomical compartment lying deep to the zygomaticus major. A greater part of the buccal space is filled by the buccal pad of fat. The other contents include the Stenson's duct, facial and buccal arteries, facial vein and branches of facial and mandibular nerves. Buccal space also has minor salivary glands and accessory parotid glands. The accessory lobe is found in about 20% of the population within the buccal space, along the line of the Stenson's duct and drains into it. Common salivary gland tumors within the buccal space are pleomorphic adenomas.

Carcinoma ex pleomorphic adenoma has an infrequent occurrence representing 3 to 5% of salivary gland neoplasm and 5 to 15% of salivary gland malignancies. It occurs most frequently in the major salivary glands especially the parotid. It occurs in recurrent or long standing pleomorphic adenoma as described by Spiro et al. The duration of the symptoms range from 1 to 48 years. The usual presentation is that of a long standing mass with sudden increase in size. The patient may also present with signs of infiltration to the surrounding structures. Invasion into the surrounding structures is an important feature in arriving at a diagnosis of malignant mixed tumor. Such tumors with malignant
areas extending beyond the tumor capsule carry a poor prognosis, the 5-year survival ranging from 25 to 65%.\(^9\) Hence they warrant an aggressive management. Our patient presented with a short history and had a well-circumscribed lesion which was unusual for a carcinoma ex pleomorphic adenoma. However, intracapsular carcinoma ex pleomorphic adenoma has been described. It is an exceedingly rare entity having a benign clinical behavior with metastatic unrecognized potential. The prognosis does not differ from that of benign mixed tumors when the malignant component is entirely contained within a fibrous capsule delimiting an otherwise benign pleomorphic adenoma.\(^9\) Although, a large series on malignant mixed tumors has reported no recurrences or metastasis in intracapsular carcinoma ex pleomorphic adenoma; there have been isolated case reports of regional metastasis from these tumors.\(^8,10\)

The usual management of patients with carcinoma ex pleomorphic adenoma involves a multimodality approach with surgical wide excision followed by adjuvant radiotherapy in view of the aggressive nature of the disease. Various surgical approaches have been described for lesions within the buccal space each with their advantages and drawbacks. The various approaches described by Rodgers and Myers et al (1988) have been tabulated (Table 1).\(^11\)

<table>
<thead>
<tr>
<th>Approach</th>
<th>Advantage</th>
<th>Drawback</th>
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<tbody>
<tr>
<td>Direct/Intraoral</td>
<td>Elimination of skin incision, limits area of tumor implantation, eliminates complication of flap necrosis</td>
<td>Risk of damage to facial nerve, Stenson’s duct</td>
</tr>
<tr>
<td>Direct cutaneous</td>
<td>Limits area of tumor implantation, eliminates complication of flap necrosis</td>
<td>Visible scar, limited exposure</td>
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<tr>
<td>Preauricular</td>
<td>Provides access to lesions posterior to the buccal space</td>
<td></td>
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<tr>
<td>Submandibular</td>
<td>Good access to lower buccal space</td>
<td></td>
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<tr>
<td>Paranasal</td>
<td>Good exposure</td>
<td>Facial nerve identification and dissection difficult</td>
</tr>
<tr>
<td>Preauricular–suborbital</td>
<td>Good exposure for addressing the neck, minimizes risk of injury to facial nerve and Stenson’s duct</td>
<td>Visible scar</td>
</tr>
<tr>
<td>Parotid–submandibular</td>
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Large case series or randomized controlled trials would be difficult to perform.

**SUMMARY**

- Buccal space lesions are a diagnostic and therapeutic challenge.
- Intracapsular carcinoma ex pleomorphic adenoma is a rare entity especially within the buccal space.
- It occurs either in the accessory parotid gland or the minor salivary glands within the buccal space.
- It has a benign course with unrecognized malignant potential.
- There are no management guidelines for intracapsular carcinoma ex pleomorphic adenoma. Large case series and longer follow-ups are required to establish definitive management guidelines.

**REFERENCES**


