

A Rare Case of Basal Cell Adenocarcinoma of Buccal Minor Salivary Gland: A Report

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Case Report

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Abstract: Basal cell adenocarcinoma (BCAC) of salivary gland is rare low-grade carcinoma, accounting for 1.6% of salivary gland tumors, tends to affect the major salivary glands. Its incidence in minor salivary gland is extremely rare, with palate being the most common site of its occurrence. To our knowledge only 72 cases of minor salivary gland BCAC have been reported. In which only 13 cases were reported from buccal mucosa. Here we report a rare case of BCAC of minor salivary gland arising from buccal mucosa. A 36 years female presented with swelling in mucosal surface of left cheek for one year. Biopsy was suggestive of basal cell adenoma. MRI showed lesion in left buccal mucosa close to mandible with no intra oral extension. Wide local excision was done. The HPE revealed basal cell adenocarcinoma of buccal mucosa and tumor margins were negative. The patient had uneventful postoperative recovery and was subjected to adjuvant radiotherapy. BCAC are low grade, slow growing, locally destructive, tends to recur and rarely metastasizing tumour. Wide local excision is the primary treatment. Neck dissection is recommended only in the presence of definitive lymphadenopathy. BCAC usually have favourable prognosis because of its low-grade nature and low metastatic potential.

Keywords: Basal cell adenocarcinoma, Minor salivary gland tumors, maxillofacial tumors, Head and neck tumors.

INTRODUCTION

Basal cell adenocarcinoma (BCAC) of salivary glands is rare low-grade carcinomas, accounting for 1.6% of salivary gland tumors, tends to affect the major salivary glands [1].

Its incidence in minor salivary gland is extremely rare, with palate being the most common site of its occurrence [1]. BCAC are locally destructive, tend to recur and only occasionally metastasizing tumor. To our knowledge only 72 cases minor salivary gland BCAC have been reported. In which only 13 cases were reported from buccal mucosa [1]. Here we report a rare case of basal cell adenocarcinoma of minor salivary gland arising from buccal mucosa.

CASE REPORT

A 36 years old female complained of swelling in the mucosal surface of left cheek for a period of one year. It was insidious in onset and gradually increasing in size. It was painless and was not associated with any fever or trauma. There was no history of any chronic disease. Patient was not a smoker and not a tobacco chewer. On inspection, there was 2×2 cm size swelling present on mucosal surface of left cheek with approximately 2 cm away from angle of mouth. Swelling was present 1cm away from lower gingivolabial sulcus and 2 cm away from upper

gingivolabial sulcus. Clinically it was not fixed to underlying muscle or mandible and was firm in consistency. Her oral hygiene was good with no loose teeth or artificial denture. No lymph nodes were

palpable. Biopsy was done outside which showed basaloid cells consistent with findings of basal cell adenoma.



Fig-01: 2x2 cm proliferative lesion in buccal mucosa

Contrast enhanced MRI of head and neck was done which showed 2.5x2.0x2.1 cm size hyperintense lesion in left buccal mucosa close to mandible with no

intra oral extension. Mild overlying soft tissue edema was seen. There were also few sub centimetric submental and submandibular lymph nodes.

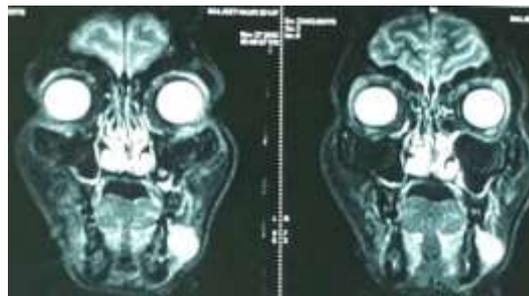


Fig-02: MRI showing hyperintense lesion in left buccal mucosa

Wide local excision was done by taking 1cm margin on all sides which showed tumor of 2.8x1.7x0.8 cm size. On HPE, tumors were arranged in clusters with peripheral pallasading. The individual cells were basaloid with high nuclear:cytoplasmic ratio and scanty cytoplasm. IHC showed positive p63 and ki 67 index of

40%. The histological picture was consistent with BCAC of cheek (buccal mucosa) and tumor margins were negative. The raw area was covered with split skin graft from thigh. Patient had uneventful postoperative recovery and patient had undergone radiotherapy for ipsilateral cervical lymph nodes.

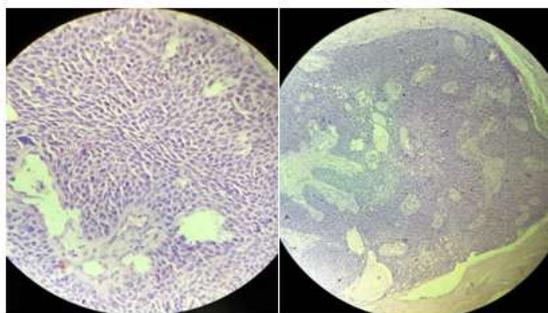


Fig-03: HPE showing features of basal cell adenocarcinoma

DISCUSSIONS

Basal cell adenocarcinoma (BCAC) was previously known under variety of names including

carcinoma ex monomorphic adenoma, malignant basal cell adenoma and basaloid salivary carcinoma. The term BCAC was officially introduced in the 2nd edition of

WHO classification of salivary tumors in 1991 and was more specifically defined in 2005 as “an epithelial neoplasm that has cytological characteristics of basal cell adenoma (BCA), but a morphologic growth pattern indicative of malignancy” [2]. It accounts for 1.6% of all salivary neoplasms and 2.9% of all malignant salivary neoplasms [1]. BCAC is a rare salivary gland tumour most commonly occurs in parotid followed by submandibular gland. Its presence in minor salivary gland is extremely rare [3]. Most common site of involvement of BCAC of minor salivary gland is palate followed by buccal mucosa, but any part of the oral cavity with minor salivary gland can get affected [1].

BCAC are low grade, slow growing tumours and most of the patients are asymptomatic. BCAC mostly involves age group of more than 60 years without any gender preponderance [4]. These tumors are locally destructive, tends to recur and rarely metastasize. The histopathogenesis of BCAC is not well established. Most tumors are thought to arise de novo, although origin from a pre-existing basal cell adenoma is possible [5].

Grossly, BCAC appears as a solid gray-tan mass and has been reported as large as 7 cm in diameter in the major salivary glands. The average lesions size was 2.4 cm (range, 0.7-4.2 cm) [1]. Histologically, BCAC are composed of basaloid cells with two morphologic appearances:

- Smaller cells with scant cytoplasm and dark nuclei.
- Polygonal cells with eosinophilic to amphophilic cytoplasm and pale basophilic nuclei.

The cells are usually arranged in nests of varying size embedded in collagenous stroma with a pallisading pattern of smaller cells at the periphery of the nests. Four histomorphological architectural patterns (solid, membranous, tubular and trabecular) have been described. The most common pattern is solid and it has high metastatic potential [4].

Immunohistochemically, BCAC's are uniformly positive for cytokeratin. Positivity for carcinoembryonic antigen and epithelial membrane antigen has been noted in the majority of the cases. These immunohistochemically profiles vary widely among the cases and none of them are found useful for confirmation of definitive diagnosis of BCAC [6].

Understanding the histology of BCAC is essential to differentiate it from other mimicking conditions like basal cell adenoma, adenoid cystic carcinoma and basaloid squamous cell carcinoma because the prognosis differs among these conditions. For example, while BCAC has a 10-year survival rate well above 75% owing to its low-grade behaviour and

low metastatic rate, the 10-year survival rate for ACC is as low as 29% [7].

BCAC shares many histologic characteristics with basal cell adenoma and mostly distinguished from basal cell adenoma by the presence of vascular or perineural involvement. Cellular atypia, presence of mitotic figures, expression of p53, p63, bcl-2 are greater in BCAC than in basal cell adenoma [4]. Adenoid cystic carcinoma is differentiated from BCAC by the presence of very high mitotic index with necrosis and pale cells with irregular angulated nuclei [8]. Unlike BCAC, basaloid squamous cell carcinoma shows squamous differentiation, higher histological grade and extensive central necrosis. High grade neuroendocrine tumors can be differentiated from BCAC by immunohistochemically examination of neuroendocrine markers.

Surgical excision with a wide margin is considered as primary treatment for BCAC [9]. Minor salivary gland tumors have a tendency for infiltration of adjacent soft tissues and therefore require wide local excision to ensure complete resection. Frozen sections can be performed to ensure the complete excision of the tumour. Regional lymph node neck dissection is recommended only in the presence of definitive lymphadenopathy [9]. Radiotherapy has been proposed for diffusely infiltrative tumours or those with perineural or vascular involvement [10]. The guidelines for metastatic disease have not been properly framed yet. Watchful waiting, radiation therapy, chemotherapy and palliative surgery would be the treatment options. Its low-grade nature and low metastatic potential makes BCAC to have a favourable prognosis.

CONCLUSION

BCAC is rare malignant lesion of salivary gland (<2% of all salivary gland neoplasms). 90% are found in parotid gland, and remainder in submandibular glands, with very few reported in minor salivary gland. This case is reported to highlight the importance about minor salivary gland basal cell adenocarcinoma and its presentation. The incidence of this tumor is rare, proper evaluation and follow up of the patient will provide more data about this tumor, thereby help the medical curriculum to provide solid information for the future generations.

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