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Indian Cancer Awareness Journal

Case Report

Acinic Cell Carcinoma of Buccal Mucosa Masquerading as Infratemporal Fossa Tumour: A Rare Case Report

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Received : 06 September 2021
Accepted : 27 September 2021
Published : 23 March 2022

DOI
10.25259/ICAJ_3_2021

Quick Response Code:

ABSTRACT

Acinic cell carcinoma (ACC) is a malignant epithelial salivary gland tumour. ACC commonly affects the parotid gland in 90% of cases. ACC rarely affects the minor salivary glands, but once affected it shows a less aggressive course. The purpose of this report is to present a rare case of ACC on minor salivary gland with an unusual presentation in an Indian male patient who was treated in our hospital.

Keywords: Acinic cell carcinoma, Minor salivary glands, Head-and-neck tumours, Low grade

INTRODUCTION

Acinic cell carcinoma (ACC) is a low-grade malignant salivary gland tumour in which some of the neoplastic cells show serous acinar differentiation with the presence of secretory zymogen granules in the cytoplasm. Acinic cell neoplasms are soft to rubbery, encapsulated, grey to pink in colour with a well-demarcated margin. Nasse in the year 1892 described tumours of the parotid gland composed of normal acinic cells, therefore, he is the first one to report the existence of an ACC.[1] Approximately 70 years ago, its malignant potential was first recognised by Godwin *et al.*[2] Although the incidence of ACC in major salivary glands range from 2.5 to 7%, their occurrence in minor salivary gland sites is rare.[3] Parotid gland is the most common site accounting for 90% of ACC, followed by intraoral minor salivary glands which account for 17–20%. Only 4% involve the submandibular gland and less than 1% the sublingual gland.[4] The tumour is usually seen in the population between 50 and 60 years of age, more commonly women (M: F ratio 1:3).[5] Here, we report a case of ACC of minor salivary gland of buccal mucosa with an unusual presentation in an Indian middle-aged patient.

CASE REPORT

A 63-year-old male presented with a swelling left side face in the cheek region of 15 years duration. Initially, the swelling was peanut size and painless, which gradually increased in size over previous 3 months. There was a dull aching pain in the region of the swelling. On examination – the swelling was 4 × 3 cm in size, hard in consistency present in the left cheek. Intraoral examination on inspection did not reveal any swelling or ulcerative/proliferative lesion on the left buccal mucosa. However, on bidigital palpation through the upper vestibule, it was found that the swelling extended – anteriorly up to level of the left upper 1st molar, posteriorly till the retromolar

trigone, superior limits could not be defined intraorally and inferiorly 2.5 cm above the lower gingivobuccal sulcus. Contrast-enhanced computed tomography base of skull to root of neck showed enhancing soft-tissue mass lesion in anterior infratemporal fossa (ITF) abutting the posterior aspect of upper buccal mucosa. No bony erosion was noted. Contrast-enhanced magnetic resonance imaging of this region was suggestive of 3.8 × 3.3 × 2.1 cm well-defined mass with lobulated outlines predominantly external to buccal mucosa overlying the left upper molars, invading the anteromedial aspect of the left masseter up to the origin of zygomatic arch and also extending into retrozygomatic and left maxillary subcutaneous fat extending into the ITF [Figure 1]. In post-contrast images, mass had mild and diffuse enhancement with no internal necrosis. One level II lymph node of size 1.1 cm was identified. No other nodes were identified in the neck. FNAC of lesion was inconclusive so a core needle biopsy was done which was suggestive of ACC. USG-guided FNAC of ipsilateral Level II lymph was reported as a reactive node. The X-ray chest of the patient was normal. Hence, a diagnosis of ACC of the left buccal mucosa was made with stage cT2N0M0. The patient underwent wide local excision of the lesion. The lesion was approached through a midline lip split incision and lower cheek flap. Intraoperatively, the lesion was found to be extending to the retromaxillary area, superiorly up to the level of zygoma and laterally abutting the coronoid process and ramus of the mandible [Figure 2]. Mass was resected with adequate margins and the lesion was sent for histopathological examination. The mucosal defect in the left buccal mucosa was reconstructed with a rotated masseter flap.

The patient had an uneventful postoperative recovery. The final histopathology report showed a well-differentiated Grade 1 ACC, size 3.1 × 2.5 × 3 cm, unifocal with all mucosal margins and base free with lymphovascular invasion positive but perineural invasion [Figure 3]. Final pathological TNM stage was $pT_2N_0M_0$. No adjuvant therapy was given. The patient showed no recurrence at 1 year follow-up.

DISCUSSION

Minor salivary gland tumours present as a heterogeneous group of tumours. The most common minor salivary gland tumours in descending order of their incidence include – mucoepidermoid carcinoma, adenoid cystic carcinoma and pleomorphic adenoma. The most common site for minor salivary gland tumour is the palate with prevalence in the range of 42–75%. Other sites include – the lips (4–21%), oral mucosa (5–16%), tongue/floor of mouth (4–12%) and retromolar area (3–7%).[6]

ACC of the salivary glands is distinctive cancers, 90% of which are low-grade tumours, however, few of them can have aggressive behaviour.[7] ACCs present as solitary firm masses that grow painlessly, most commonly in the parotid gland, rarely in the minor salivary glands. They show various histological patterns which include – solid, microcystic,

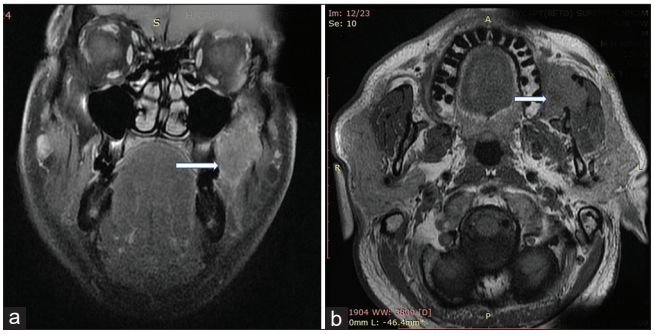

Figure 1: CEMRI base of skull to root of neck, a. T2W coronal cut showing left side lesion external to upper buccal mucosa extending to ITF (white arrow), b. T1W axial cut – left side same lesion (white arrow).

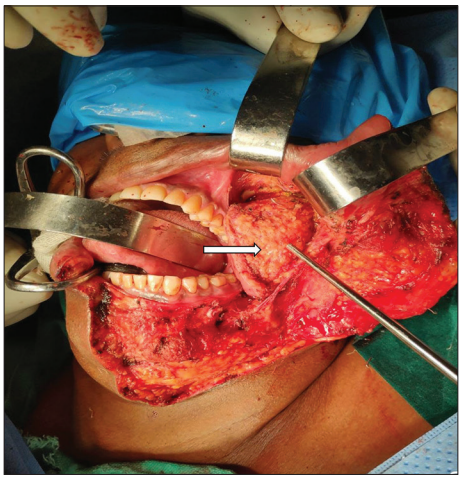

Figure 2: Intraoperative picture – lower cheek flap after midline lip-split incision, mass extending from buccal mucosa into ITF (white arrow).

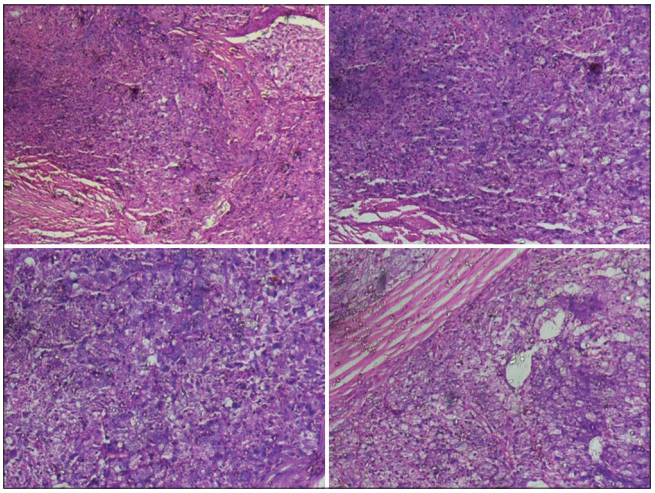

Figure 3: Histopathology slides of the final resection specimen showing ACC.

papillary-cystic and follicular.[8] Histologically, we see serous acinar cells, with an eccentric nucleus darkly stained with abundant granular basophilic cytoplasm having coarse granules, admixed with clear, vacuolated and intercalated ductal cells. Due to the basophilic cytoplasm, they are called 'blue dot tumours.' Other lesions which mimic ACC are sialadenosis, Warthin's tumour, oncocytoma, granular cell tumour, basal cell adenoma and pleomorphic adenoma.[9] Hence, it can be a diagnostic challenge for pathologists.

ACC has a tendency for recurrence. ACC of the minor salivary glands rarely show metastasis. Metastases may occur to the cervical lymph nodes by lymphatic spread and hematogenously to the lungs or bones.

Complete surgical excision of the tumour by intraoral or extraoral approach is the treatment modality of choice depending on the location and spread of the tumour. Elective neck dissection is not done as the regional metastases are <10%. Well-encapsulated lesions that are removed in toto have less chance for recurrence. Radiotherapy is indicated in unresectable tumours or in adjuvant settings post-surgery when there are high-risk factors such as high stage or high grade. Chemotherapy for ACC is considered ineffective, except for pain relief or partial response. ACC has a good prognosis with 5 years overall survival rate (OSR) of >95% and 20 years OSR of >85%.[10] Long-term follow-up of these lesions is necessary to document the success of treatment. Awareness about the unusual presentations as seen in our case (appeared to be an ITF mass), behaviour and prognosis of this tumour is important for optimal management of these patients.

CONCLUSION

This case has been reported as it is a rare presentation of ACC of buccal mucosa masquerding as an infratemporal fossa mass lesion. Cinicians should consider ACC of buccal mucosa too in the differential diagnosis of such masses.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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How to cite this article: Dwivedi G, Gujrathi S, Sood A. Acinic cell carcinoma of buccal mucosa masquerading as infratemporal fossa tumour: A rare case report. Indian Cancer Awareness J 2022;1:21-3.