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# **An Unusual Adrenal Gland Metastasis in a Case of Squamous Cell Carcinoma of Buccal Mucosa**

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### **Authors' contributions**

*This work was carried out in collaboration between all authors. All authors read and approved the final manuscript.*

### **Article Information**

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### **Case Study**

## **ABSTRACT**

The majority of oral cavity cancers presents as localized disease and remain localized until late in the course of their development. Distant metastasis is very rare in carcinoma of buccal mucosa and has a very dismal prognosis. Distant spread is either related to uncontrolled local disease or inadequate local treatment. Lungs and bones are common site for metastases. Extensive metastatic workup should be considered in locally advanced buccal mucosa patients, who either defaulted complete treatment or have an uncontrolled loco-regional relapse.

We report a case of unusual adrenal metastasis from Squamous Cell Carcinoma (SCC) of buccal mucosa. 42 years old lady initially presented with cT2N1M0 SCC of buccal mucosa (left) underwent radical surgery followed by post-operative Radiotherapy. After eight months of follow up she developed local relapse. After an initial response to salvage systemic chemotherapy, patient discontinued chemotherapy. After 6 months of discontinuation patient had gradually increasing abdominal pain. Contrast Enhanced Computer Tomography Scan (CECT) showed Left adrenal mass with para-aortic lymph node. Other investigations were normal. CT guided Fine needle Aspiration Cytology (FNAC) showed metastatic deposit of squamous cell origin. Patient is being treated with Tab Gefitinib and supportive medications.

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## 1. INTRODUCTION

Carcinoma of buccal mucosa is the most common carcinoma of the oral cavity in Southeast Asia because of the widespread use of betel nut [1]. The majority of oral cavity cancers presents as localized disease and remain localized until late in the course of their development. Distant metastasis occurs in approximately 15% to 20% of patients who eventually die of their disease [2]. In general terms with respect to head and neck cancer, 66% of distant metastases are to the lungs, 22% to the bones, and 9.5% to the liver [3]. Rarely other sites of distant metastases have been reported; including brain, pericardial cavity, liver, lung, thyroid, bone and bone marrow. Suhag V et al. reported first case of adrenal gland metastasis reported in 2011 [4]. We report probably the second case of adrenal metastasis in advanced buccal mucosa till date.

## 2. CASE REPORT

A 42 years old woman without any co-morbidity presented with ulcero-proliferative growth at left gingiva-buccal sulcus extending to left retro-molar trigone along with single ipsi-lateral neck node at level Ib. After work up she was staged as cT2N1M0, Carcinoma buccal mucosa (Left) (Figs.1, 2). She underwent wide local excision with segmental mandibulectomy with modified radical neck dissection (left) followed by Pectoralis myo-cutaneous flap reconstruction on 30/1/2015. Staging was pT2N2M0 (SCC).Depth was 8mm along with lympho-vascular space invasion. Patient was treated with Concurrent chemo-radiation (with weekly Inj. Cisplatin) from 13/4/2015 to 2/6/ 2015. She had a local relapse along with nodal relapse in February 2016. (rCT2N2bM0). She underwent weekly Inj. Cetuximab, Inj. Paclitaxel and Injection Carboplatin from 5/4/2016 to 11/06/2016. (total 6 weeks). On evaluation in June 2016 she had Partial Response. After that Patient discontinued chemotherapy. In December 2016, she attended our Out Patient Department (OPD) with intractable abdominal Pain. Ultrasonography (USG) abdomen showed Left adrenal mass along with enlarged Para-aortic node. CECT abdomen showed (29/12/2016) 3X3X2.5 cm soft tissue mass in Left suprarenal gland along with centrally necrotic lymph node in retro-peritoneum. Guided FNAC showed a few clusters of malignant squamous cells with

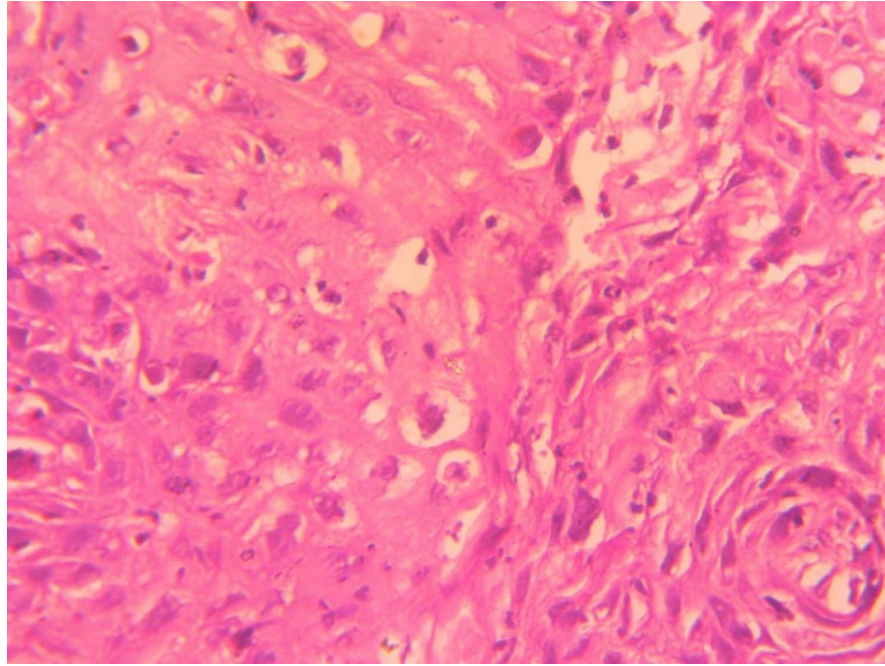
nuclear hyperchromasia, anisonucleosis and caudate shaped cytoplasm. Background shows scattered mature squamous cells, enucleate squamous and necrotic debris, consistent with metastatic deposit of SCC (Figs. 3, 4). Patient had no evidence of disease at head neck region. Gynecological examination finding was normal. CECT Thorax was normal. Colonoscopy was normal. Serum CEA was within normal range. Patient is being treated with oral Gefitinib tablets along with supportive medication.

## 3. DISCUSSION

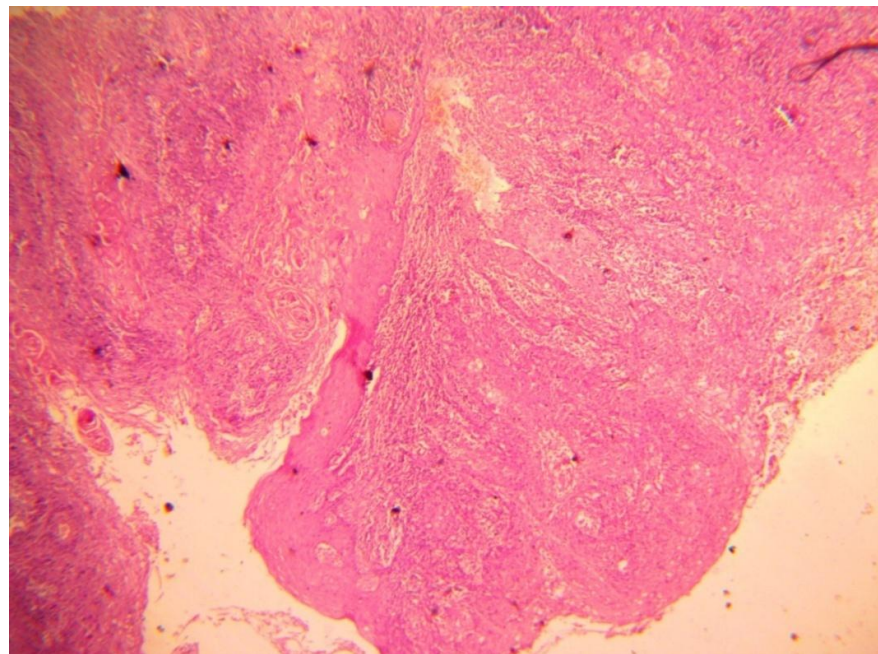
SCC of the oral cavity ranks as the twelfth most common cancer in the world and the eighth most frequent in males. The majority of oral cavity cancers presents as localized disease and remain localized until late in the course of their development. Distant metastasis occurs in approximately 15% to 20% of patients who eventually die of their disease. However, SCC of buccal mucosa is the most common oral cancer in men and the third most common oral cancer in women in India; and accounts for up to one-third of all tobacco-related cancers [4,5]. The patient may present with pain, bleeding, trismus and cervical lymphadenopathy. In advance cases, tumor may destroy the entire cheek and invade bone. Carcinoma buccal mucosa most commonly spread directly spread to adjacent organs e.g. gingiva-buccal sulcus, alveolar ridge, hard palate. Lymph node metastasis occurs in 9-30% patients during the course of treatment [6]. In oral cavity cancer, local recurrence is more common than regional recurrence, and has been reported between 23- 32% [4].

Distant metastasis is very rare, as patient often die due to uncontrolled local disease before distant metastases manifest clinically. Neck node metastasis, extra-capsular spread and lympho-vascular space invasion increase the risk for distant metastases. Distant metastases generally have poor prognosis, with ninety percent mortality within 1 to 16 month (median 3 months) of the evidence of distant metastasis [4]. Distant metastases typically manifest themselves in the lung, bones, liver and skin. There are sporadically reported cases worldwide of atypical presentations of distant metastasis in head neck SCC. Very rarely it occurs in carpal bones, limb muscles and cardiac tissues [7,8]. Metastasis to adrenal gland is very unusual. Only one case has

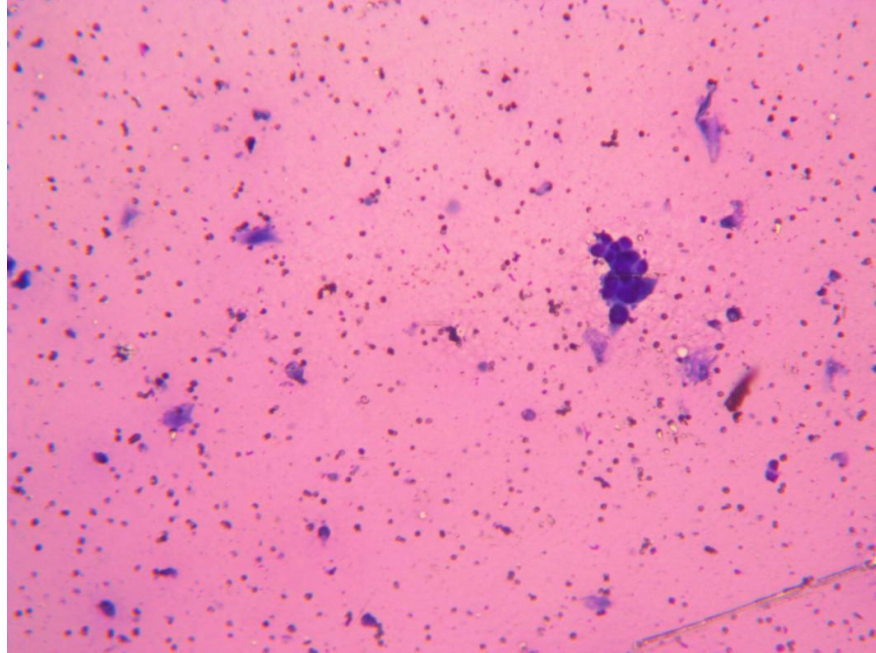
been reported till today. Suhag V et al. reported case of locally advanced SCC of buccal synchronous metastasis of adrenal gland in a mucosa [4].



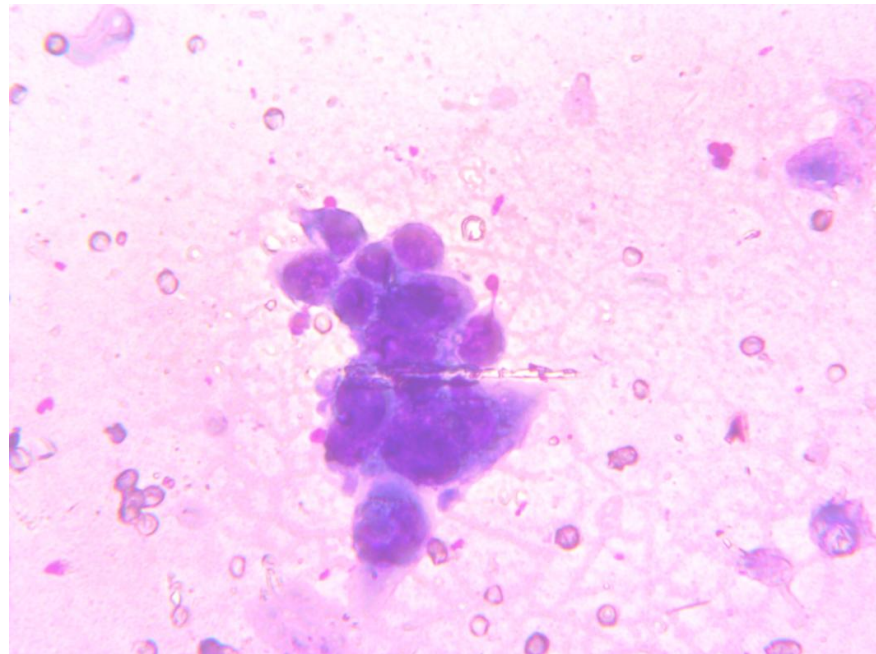
**Fig. 1. Biopsy smears from left buccal mucosa showing clusters of malignant squamous cells (10 X 40 view of malignant cells in inset)**



**Fig. 2. Biopsy smears from Left Buccal Mucosa showing clusters of malignant squamous cells (10 X 10 view of malignant cells view in inset)**



**Fig. 3. FNAC smears from adrenal lesion showing clusters of malignant squamous cells (10 X 10 view of malignant cells view in inset)**



**Fig. 4. FNAC smears from adrenal lesion showing clusters of malignant squamous cells (40 X 10 view of malignant cells view in inset)**

Our reporting is second case with adrenal metastasis and first case as metachronous adrenal deposit. Majority of buccal mucosa fails locally, of which 90% occurs within 1-2 years of

diagnosis. Distant metastasis is rarest among all head neck sub sites [4]. In our case, the patient had a local relapse after eight months of follow up. The patient initially responded to salvage

chemotherapy, which she discontinued. Subsequently she had adrenal metastasis within one year of salvage treatment.

#### 4. CONCLUSION

Distant metastasis is rare in SCC of buccal mucosa with dismal outcome. Adrenal metastasis is very unusual. Extensive metastatic workup should be considered in locally advanced buccal mucosa patients, who either defaulted complete treatment or have an uncontrolled loco-regional relapse.

#### CONSENT

All authors declare that written informed consent was obtained from the patient (or other approved parties) for publication of this paper and accompanying images.

#### ETHICAL APPROVAL

All authors hereby declare that all experiments have been examined and approved by the appropriate ethics committee and have therefore been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki.

#### COMPETING INTERESTS

Authors have declared that no competing interests exist.

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