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Original Article

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Small Cell Carcinoma of Oral Cavity (Buccal Mucosa): A Rare Case Report

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Abstract

Small cell carcinoma (SCC) is an exquisite head and neck cancer defined as a high-grade neuroendocrine tumor analogous to a Small Cell Lung Carcinoma (SCLC). In the head and neck region, it is commonly located in the larynx, followed by the sino-nasal tract, the salivary glands, the trachea, the oral cavity, and the oropharynx in descending order. To the best of our knowledge, limited case reports have been documented up to now with a questionable definitive treatment protocol. We report the fourth case of SCC of buccal mucosa, which was confirmed by histopathology and immunohistochemistry and a wide local excision performed.

Keywords: Small cell carcinoma, Oral cavity, Buccal mucosa

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Introduction

Small cell carcinoma (SCC) is the most frequent and aggressive lung malignancy and has a propensity for both local and distant metastasis. [1] There is a preponderance to the pulmonary origin. However, they can also originate from extra-pulmonary sites. Amongst them, 2.5-5% arise outside the lung, of which only 11-16% arise from the head and neck region. [2] The commonest site for extra-pulmonary SCC is the larynx. The tongue, salivary glands, esophagus, trachea, and paranasal sinuses are some of the additional locations. [3] SCC of the oral cavity is exceptionally rare. To the best of our knowledge, only 3 cases of SCC of the buccal mucosa were documented.[4,5,6] We now address the fourth case of small cell carcinoma of buccal mucosa and the first case reported in a female patient.

Case Report

A 72-year-old female presented to ENT OPD with complaints of pain and swelling in the left cheek for the last three months. She also gave a history of heavy chronic smoking (1 bundle/day) for more than 40 years. She had undergone a biopsy elsewhere, which suggested poorly differentiated squamous cell carcinoma.

On physical examination, a 1*2 cm smooth globular pinkish red tender firm mass arising from the left buccal mucosa extending superiorly at the level of upper alveolus to 1 cm below it, just anterior to the ramus of the mandible and extending anteriorly up to the last molar was seen [Figure1]. There was no clinically palpable cervical lymphadenopathy. On slide review in our institute, poorly differentiated malignancy was reported, and immunohistochemistry was advised, which demonstrated positivity to cytokeratin (CK), Synaptophysin and weakly positive for chromogranin and negative for S -100, suggesting it to be a small cell carcinoma. The Contrast-enhanced computed tomography (CECT) from the base of the skull to the diaphragm showed an enhancing lesion involving left buccal mucosa without any bony involvement and cervical metastasis [Figure 2] however, distant metastasis in the lung was noted.

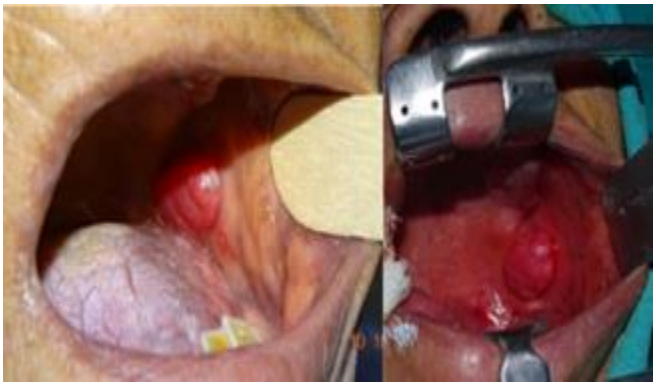

Figure 1: Preoperative (right) and intraoperative (Left) oral cavity examination

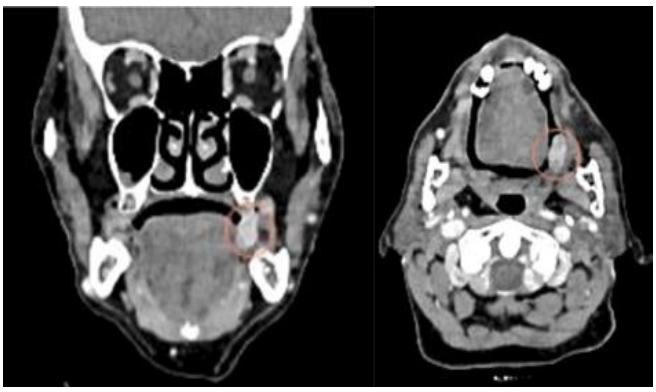

Figure 2: Coronal (right) and axial (Left) images of CECT showing contrast-enhanced lesion

Under general anaesthesia, wide local excision with adequate margins was performed trans-orally because the tumor was thought to be easily resectable. The specimen was then sent for histopathological analysis, which revealed that it was a small-cell carcinoma with clear margins. Postoperatively the patient was advised to adjuvant chemotherapy. The patient is in regular follow-up and is free of symptoms with no evidence of recurrence /residual disease.

Discussion

Head and neck SCC is an unusual clinical problem. It still needs to be determined where oral SCC originated. Oral squamous epithelium harbours neuroendocrine cells, which are thought to be the third division of the nervous system. Though it has been suggested by a few studies that these are the cells of origin, there is a strong assumption that the pluripotent cells in the squamous epithelium of the oral cavity and minor salivary glands could be the site of origin. Three subtypes of neuroendocrine carcinomas are the well-differentiated NE carcinoma, the typical carcinoid tumor, the moderately differentiated NE carcinoma or the atypical carcinoid tumor; and the poorly differentiated carcinoma or the small cell carcinoma.[7,8] SCC is also known as oat cell carcinoma, anaplastic small cell carcinoma, small cell neuroendocrine carcinoma of intermediate type and small cell neuroendocrine carcinoma.[2]

These are frequently observed in men and are usually associated with heavy tobacco smoking.[6] They are histologically analogous to small cell carcinoma of the lung.[9] SCC exhibits positivity for neuroendocrine markers such as neuron-specific enolase, synaptophysin, chromogranin A and neurofilament.[10]

Due to the oddity of these tumors, the evidence of treatment recommendations and clinical outcomes is not abundant. Recently in June 2017, the University of Wisconsin School of Medicine and Public Health conducted the most extensive retrospective study on SCC of head and neck region adopting the National Cancer Database (NCDB). According to this study, the median overall survival was 36.4 months for the nasal cavity primary tumor, 23.7 months for the oropharynx, 20.8 months for the oral cavity, 17.9 months for larynx/hypopharynx, 15.1 months for nasopharynx with best median overall survival for nasal cavity and paranasal sinuses tumors and the worst being nasopharynx. The overall survival, according to this study, was 20.3 months. Combined chemoradiotherapy was the frequently given treatment modality. Amongst the stage I/II patients, no overall survival difference with treatment type was noted. In locally advanced diseases, surgery added no benefit to chemoradiotherapy. In metastatic disease, chemotherapy alone had a prolonged survival, and the addition of radiotherapy did not improve the survival.[11]

Chemotherapeutic drugs used in the head and neck SCC are Cisplatin and Etoposide, as used in the treatment of Lung SCC.[11]

Our patient had a localized lesion in the buccal mucosa. Therefore, excision was the first step, proceeded by adjuvant chemotherapy. After 14 months of surgery, the patient had no disease. Long-term continuous follow-up is mandatory for recognizing any local or distant recurrence.

Conclusion

An uncommon instance of small cell neuroendocrine carcinoma (SNEC) of the buccal mucosa in a female is presented in the current study. Clinicians should take into account the possibility of this aggressive tumor, especially in heavily smoking elderly patients. A patient must undergo a thorough clinical evaluation and morphological and immuno-histochemical testing to rule out a metastatic tumour. The preferred management approach for patients with localised lesions may be surgery combined with adjuvant chemotherapy. However, the conventional treatment plan for patients with oral SNEC is still unknown due to the paucity of data.

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