Case Report

Bilateral squamous cell carcinoma of buccal mucosa in a young adult man: A case presentation with review of literature

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Abstract

The control of oral squamous cell carcinoma (SCC) is difficult even after treatment because it has a tendency to multiple primary carcinomas. The incidence of second primary neoplasm in the head and neck region cannot be forecasted reliably. The purpose of this report is to describe the clinicopathological characteristics of a unique case of bilateral buccal mucosal SCC in a 35-year-old male and to determine whether any associated risk factors are present.

Keywords: Bilateral, buccal mucosa, squamous cell carcinoma

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INTRODUCTION

The control of oral squamous cell carcinoma (SCC) is difficult even after treatment because of its great tendency for multiple primary carcinomas (MPCs). The epidemiological information on oral MPCs is limited.[1] The propensity for the development of multiple mucosal cancers is offered to result from field cancerization, which creates a diffuse field of altered epithelial cells with increased potential for malignant change.[2] MPCs can develop simultaneous (diagnosed at the same time as the primary tumor), synchronous (identified within 6 months of the primary tumor) or metachronous (occur more than 6 months of the primary tumor).[3] Oral MPCs in patients without tobacco or alcohol abuse have the next occurrence only in the oral cavity. The gingiva is the most common site of occurrence.[1,4] Here, we describe the clinicopathological characteristics of a case of bilateral buccal mucosal SCC in a young adult man and discuss about its associate risk factors.

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CASE REPORT

A 35-year-old male was referred to a private oral pathology center for the evaluation of a painless right buccal mucosal mass with 4–5 months' duration. The patient's past medical history indicated type 2 diabetes mellitus (fasting blood sugar = 300 mg/dl) and positive hepatitis B surface antigen (which had been unknown to the patient prior to his blood transfusion). He also denied any use of alcohol and cigarettes. The patient pointed out that his father was an addict and died because of laryngeal cancer. His mother and brother were also affected with hepatitis

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B. The patient was a farmer and had no other close relationship to harmful chemical or industrial substances. In the clinical examination of the right buccal mucosa, a nontender mass, measuring 5.5 cm × 5 cm with a verruciform surface, was identified [Figure 1]. The lesion had a soft-to-firm consistency. There was no evidence of the neck lymphadenopathy. An incisional biopsy was performed under local anesthesia. Microscopic sections of the lesion showed sheets of malignant squamous epithelium invading into the lamina propria. The epithelial cells showed pleomorphism, hyperchromatism and remarkable mitotic features without keratin pearl formation [Figure 2]. According to these features, the diagnosis of SCC was made, and immunohistochemical staining for ki-67 and P16 (in order to rule out the human papillomavirus [HPV] infection) was done. The cells were negative for P16, but nearly 100% of the cells were positive for ki-67 [Figure 3]. The whole-body scan did not detect any distant metastases.

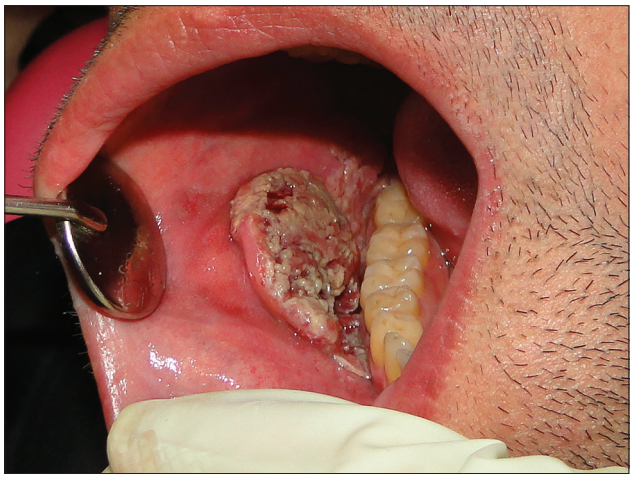

Figure 1: A nontender mass with a verruciform surface was present on the right buccal mucosa. The lesion had a soft-to-firm consistency

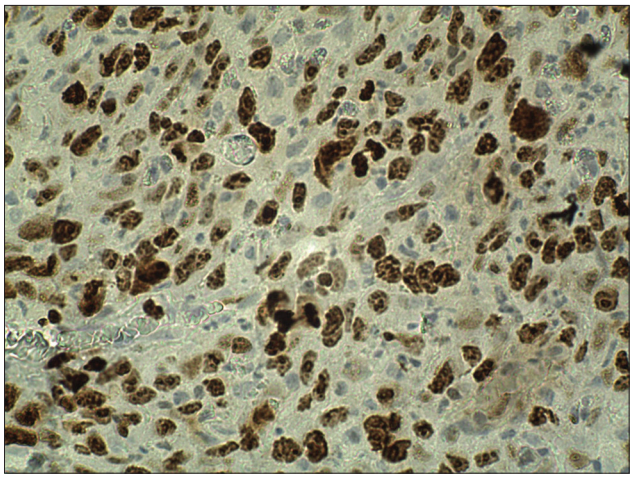

Figure 3: Ki-67 is highly expressed as brown nuclei in dysplastic sheets and islands (immunohistochemistry ×400)

According to these data, clinical staging was reported as T3N0M0 (Stage III), and the patient was referred to a cancer institute for treatment. Radical surgery (with cutaneous graft), modified radical neck dissection, 40 sessions of radiotherapy and 8 sessions of chemotherapy (5-FU plus cisplatinum regimen) were prescribed. The patient was under follow-up until 18 months when he came back with an elevated white plaque lesion on his left buccal mucosa measuring 1.5 cm × 1.2 cm × 0.5 cm [Figure 4]. The excisional biopsy with safe margins was done, and microscopic evaluation revealed a well-differentiated SCC. He is now well after 3-year follow-up, and there is no clinical evidences of recurrence or new lesion.

DISCUSSION

Warren and Gates[5] described a set of diagnostic criteria for "MPCs" as follows: (a) the two neoplasms must be malignant, (b) the two neoplasms must be anatomically

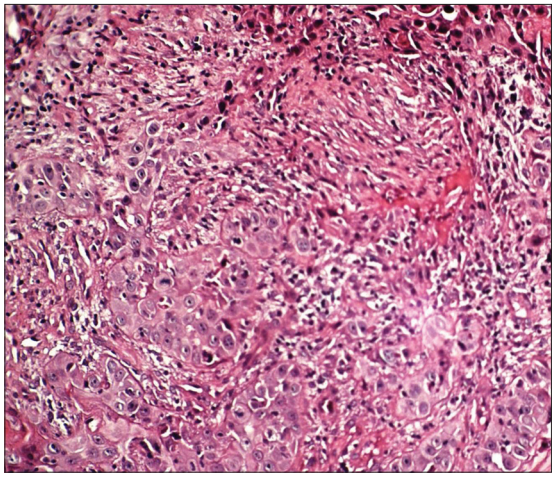

Figure 2: Microscopic sections revealed malignant squamous sheets and nests in the underlying connective tissue (H and E, ×100)

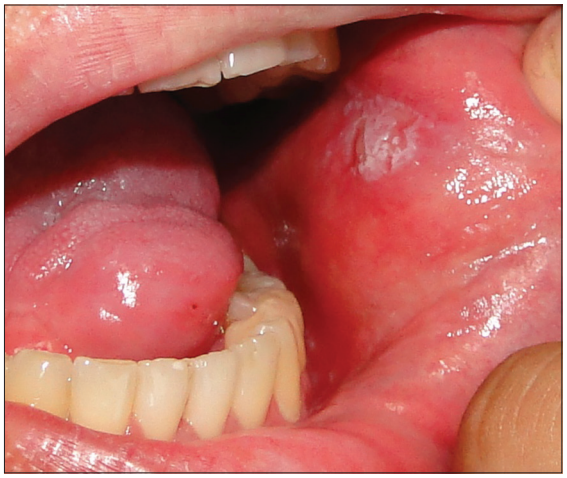

Figure 4: An elevated white plaque lesion on his left buccal mucosa

separated and not connected by epithelial or submucosal neoplastic changes, and (c) the possibility of metastasis from the primary tumor must be eliminated. All these criteria were found in the present case. MPC has a tendency to occur in females.[1] The most common site of involvement is the gingiva followed by the tongue,[4] which is inconsistent with our case. Lai *et al*.[6] found that second primary cancers in Taiwan are common in male patients who are affected with SCC of buccal mucosa. They mentioned that one of the major risk factors for SCC of buccal mucosa in Taiwan is chewing betel quid. Researchers have found that the second primary tumor can reveal some or all genetic markers of the primary tumor. This indicates that both neoplasms have originated from one solitary preneoplastic field with common clonal progenitor cells.[7] Patients with dyskeratosis congenital and Fanconi anemia have a tendency to develop oral SCC in younger age groups.[8] Our patient did not have any sign and symptoms of these syndromes. The present case had a poorly controlled diabetes type 2 and poor glycemic control. Obesity, a diet rich in fat and diabetes, may increase the risk of oral SCC.[9] Thomson[10] found that abnormal microscopic changes could occur in clinically normal-looking mucosa at corresponding anatomical sites. Here, the patient was suffering from hepatitis B. Some investigators suggest that hepatitis C virus infection is strongly related with the existence of MPCs.[11] However, this association has not been stated with hepatitis B. Field cancerization does not appear to be associated with malignancies attributed to HPV infection.[2]

A history of proliferative verrucous leukoplakia and oral lichen planus have also been mentioned in MPCs.[1,4] Elevated levels of heavy metal pollutants such as nickel, chromium and arsenic in farm soil and increased blood concentrations of some of these metals in affected patients have been described. As well, an elevated risk of oral cancer is reported for workers in the wood product industries who are chronically exposed to certain chemicals, such as phenoxyacetic acids.[2]

No reliable method has been yet developed to estimate which of the oral SCC patients will progress second primary tumors.[12] Some investigators suggest that the biologic behavior of MPCs is different from single carcinomas.[1,6] Multiple cancers generally have a poor prognosis.[3] In contrast, Mochizuki *et al*.[1] stated that MPCs are less likely to occur as advanced disease or with metastasis of cervical lymph nodes. In our patient, lymph node involvement was not seen; however, the tumor stage was III. It has been suggested that external beam radiotherapy may be related with a reduced incidence of second malignancy for localized oral cancer.[13] Nevertheless, the second neoplasm developed in our patient despite chemoradiotherapy. Second MPCs may develop long after onset of the first carcinoma.[1] Therefore, oral SCC patients need a long-standing and close follow-up due to the occurrence of second primary tumors.[12] In conclusion, because of the possibility of developing a second cancer in patients with oral SCC, periodic examination of whole oral cavity mucosa as well as the original location of the tumor is recommended. On the other hand, as diabetic patients are immunocompromised, they should have periodic examination of the oral mucosa and the blood sugar level must be well controlled. Furthermore, in all patients with oral SCC, especially nonsmokers and nondrinkers, associated underlying causes such as employment, nutrition and infection must be carefully investigated.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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