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Contemporaneous Bilateral Presentation of Oral Primary Carcinoma - A Sporadic Case Report

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Abstract: ***Introduction: The incidence of oral squamous cell carcinoma is increasing in developing countries due to the over - abuse of tobacco and stressful lifestyle. Management of single primary oral carcinoma has been challenging, due to its characteristics of lymphatic spread and loco - regional recurrence even after 3 tier oncological management with surgery, radiation therapy and chemotherapy. Hence multiple primary carcinomas occurring in oral cavity has its own unique presentation and characteristics, due to the incidence of two neoplasms occurring from a single preneoplastic field with common successor progenitor cells. Any primary tumor will show all or a few genetic markers of the other multiple tumors occurring in the region. Rarely contemporaneous occurrence of bilateral oral primary is seen, thus the significance of timely diagnosis, and early intervention is very crucial in deciding the prognosis. Case Presentation: Here we present a rare case of a 48 - year – old female known case controlled hypertension with a habitual history of betel nut and tobacco abuse for the last 15 years. Patient developed bilateral simultaneous buccal mucosa lesions. CT scan revealed a heterogeneous enhancing lesion involving bilateral gingivobuccal mucosa, and multiple enlarged bilateral level I, II, and left level IV lymph nodes were noted. A wide local excision of bilateral buccal mucosa with bilateral neck dissection and right hemimandibulectomy or segmental mandibulectomy was planned. Discussion: MPCs are usually not aggressive and are never found to be advanced diseases with cervical lymph node metastasis. In our case, the disease was advanced, with radiographic evidence of cervical metastatic lymph node at left level 1b. There is not much literature published about the MPCs occurring in the oral cavity and their management. Thus, it makes this case report very unique for presentation. The diagnostic technique to investigate, which primary oral SCC has occurred first in MPCS is not yet found. Many authors suggested that the characteristic behavior of single primary oral SCC over MPCS is very contrasting. Thus making the management more challenging and requires the team approach. Reliable techniques need to be found or developed to know the occurrence of the first primary in MPCS so that effective management protocol could be followed in the treatment of MPCS to benefit the patient. Conclusion: Early diagnosis and early intervention are keys to success in the treatment of all oral cancers. Similar to the MPCs too, with today's advanced imaging modalities like PET scans, MDCT scans, and MRI scans, early diagnosis is possible. Thus, bilateral primary resection in the oral cavity is surgically challenging for all surgeons, but effective surgical clearance of a 1cm margin is key to success with bilateral neck dissection and reconstruction.***

Keywords: OSCC (Oral squamous cell carcinoma), MPCs (Multiple primary carcinomas). CT (computed tomography)

1. Introduction

Worldwide, the occurrence of oral squamous cell carcinoma (OSCC) is increasing, with a significant increased risk of lowered survival rates less than 5 years. OSCC ranks 6th globally in the incidence of overall cancers diagnosed every year. [1] Oral cancers in India account for 8–10%, which contributes around 33 percent of the total oncological burden. [2] Hence, prevention of OSCC is challenging even after 3 - tier (surgery, radiotherapy, and chemotherapy) treatment of oral cancers due to the tendency for multiple primary carcinomas (MPCs). The incidence of MPCs is about 7% to 21% in the head and neck region and around 1.4% in the oral cavity alone. [2] MPCs can occur simultaneously with primary tumours and are termed synchronous if diagnosed 6 months after the occurrence of the primary tumour or metachronous if diagnosed 6 months after the occurrence of the primary tumour. [3]. Oral MPCs have the characteristic of occurring only in the oral cavity in patients without a habitual history of consumption of alcohol and tobacco. The commonest site for the occurrence of oral MPCs is the buccal mucosa and gingiva [4, 5]. Here we present a rare case of bilateral oral primary carcinoma.

2. Case Report

A 48 - year - old female reported to our department's OPD with a chief complaint of bilateral buccal mucosa lesions for the last 3 months. She was a known case of controlled hypertension with a habitual history of betel nut and tobacco chewing for the last 15 years. The patient presented with a history of simultaneous gradual increases in lesion size in the bilateral buccal mucosa. Lesions were associated with mild pain, which was aggravated by chewing hard food and

opening the mouth and relieved by taking medications. No relative history of dysphagia, odynophagia, hoarseness of voice, dyspnea, oral bleed, paresthesia of the lower lip, or any active discharge from the tumour was noted. On extraoral examination, gross facial asymmetry was noted in the right lower third of the face due to underlying well - defined swelling of the lesion, with grade 2 trismus (mouth opening of 27mm) and overlying skin that appears normal. Intraoral examination revealed an ulceroproliferative growth in relation to the right lower gingivobuccal sulcus, extending antero - posteriorly from the distal aspect of the 42 tooth to the distal aspect of the 46 molar tooth (not involving the RMT region). (Figure 1) Another ulceroproliferative lesion was noted in the left buccal mucosa, extending antero - posteriorly from mesial aspect 33 to the distal aspect of 38teeth (not involving the RMT region) and supero - inferiorly without involving the upper and lower gingivobuccal sulcus. (Figure 2) On clinical palpation left and right I B lymph nodes were palpable, left I B was measuring approximately 1.5 cm in all dimension which was fixed, stony hard in consistency. Patient underwent incisional biopsy of bilateral lesions under local anesthesia, hemostasis was achieved and both the specimens were labelled separately and was sent for histopathological examination.

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Histopathological examination: Both specimens were tissue processed and were reported as having moderately differentiated squamous cell carcinoma.

Radiological interpretation: MDCT multislice scan were performed of face, neck and thorax and brain region with contrast and showed heterogeneous enhancing lesion involving bilateral gingivobuccal mucosa. (Figure 3) And also cortical bone loss (involvement) was noted in relation to right mandible at premolar teeth region. Multiple enlarged bilateral level I, II and left level IV lymph nodes were noted, largest measuring 2.7cm x 2.0cm in left level I B with loss of fat plane with submandibular gland. (Figure 4 and 5) CT scan of the thorax, brain and usg of the abdomen did not show any evidence of distant metastasis. Hence overall pre - op clinical staging was made as T4N2cM0 stage 4A. (AJCC 8th edition and NCCN guidelines).

Surgical procedure: A wide local excision of bilateral gingivobuccal lesion was done with right side hemimandibulectomy, right side selective neck dissection was performed with level I to III clearance. Left side modified radical neck dissection type 2 was performed with level I to V lymph node clearance and reconstruction of resected primary defect was done with local flap. This case report has been reported according to the CARE 2013 criteria.

3. Discussion

Multiple primary carcinomas (MPCs) occur about 7 to 21 percent in the head and neck region and have a very low incidence of about 1.4 percent in the oral cavity. [2] And are usually seen more in the gingiva region, followed by the tongue. [4, 5, 8], which was consistent with our case. To call MPCs, they have to meet the diagnostic criteria as described by Warren and Gates. First, the two neoplasms or the lesion should be malignant; second, the two malignant neoplasms should be present anatomically at two different sites and should not be connected by epithelial or submucosal neoplastic variations; and lastly, evidence of metastasis from the primary tumour should be excluded. [6] All criteria mentioned above were present in this case.

Squamous cell carcinomas occurring in the oral cavity have an overall disease - free survival rate of 5 years. Hence, the study conducted by Ya - Dong Li et al. [7] indicates that there is not much significant variation noted in the overall survival rate of MPCs over the single primary lesions of the oral cavity. The overall survival rates reported in this study were 90.7% and 79.6% at 5 years and 10 years, respectively. Usually, MPCs are not aggressive and are never found to be advanced diseases with cervical lymph node metastasis. [9] In our case, the disease was advanced, with radiographic evidence of cervical metastatic lymph node at left level 1b.

There is not much literature published about the MPCs occurring in the oral cavity and their management. Thus, it makes this case report very unique for presentation. MPCs occurring in the oral cavity has multiple etiological and high - risk factors. These factors include long - term uses of tobacco (45%), areca nut (25%), bettle nut (14%), and alcohol (16%). [10] MPCs also has a higher incidence in patients with Fanconi anaemia, hepatitis C virus, and those who work in the wood and plastic industries and are exposed to hazardous chemicals (phenoxyacetic acid) and heavy metal pollutants such as nickel, arsenic, and chromium. [11, 12,]

The concept of field of cancerization is always associated with MPCs occurring in the oral cavity, which is secondary to tobacco abuse in any form. Where a range of morphological cell differentiation occurs in the layers of mucosa and epithelium, leading to an area of skip lesion, causing premalignancy [13]. Usually, any primary tumor will show all or a few genetic markers of the other multiple tumors occurring in the region. Thus indicating the incidence of two neoplasms occurring from a single preneoplastic field with common successor progenitor cells. [14] Our patient had a history of long - term tobacco chewing with betel nuts, which have a significantly high risk of causing oral cancer.

The diagnostic technique to investigate, which primary oral SCC has occurred first in MPCS is not yet found. [15] Many authors suggested that the characteristic behavior of single primary oral SCC over MPCS is very contrasting. [9] thus making the management more challenging and requires the team approach. Hence surgery was performed first in this case as both the primary lesion and disease were localized and were restricted to the oral cavity and neck, without any evidence of distant metastasis. Our patient also underwent post - surgical IMRT radiotherapy treatment bilaterally up to 60 GY of 30 cycles and tolerated well during the course of treatment. At 6 months follow up patient is doing well with good functional and esthetic outcomes without any signs of recurrence. Rarely contemporaneous occurrence of bilateral oral primary is seen thus the significance of timely diagnosis, and early intervention is very crucial in deciding the prognosis. Reliable techniques need to be found or

developed to know the occurrence of the first primary in MPCS so that effective management protocol could be followed in the treatment of MPCS to benefit the patient.

4. Conclusion

Early diagnosis and early intervention are keys to success in the treatment of all oral cancers. Similar to the MPCs too, with today's advanced imaging modalities like PET scans, MDCT scans, and MRI scans, early diagnosis is possible. Thus, bilateral primary resection in the oral cavity is surgically challenging for all surgeons, but effective surgical clearance of a 1cm margin is key to success with bilateral neck dissection and reconstruction. The disease - free survival rate of MPCs is similar to that of single primary cases if simultaneous postoperative adjuvant chemotherapy and radiotherapy are received. Furthermore, patients with MPCs should be screened for habit history, occupational history, and viruses to attribute the risk factors for MPCs to know the prognosis.

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Figure 1

Figure 2

Figure 5

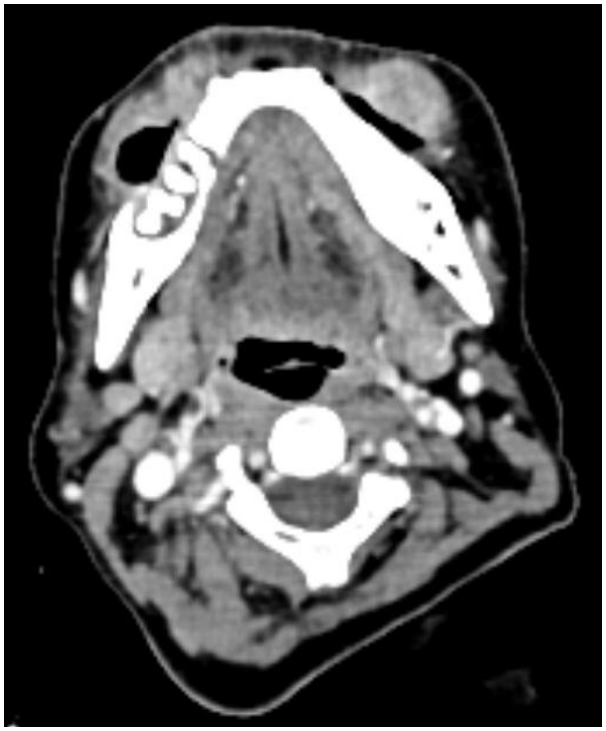

Figure 3