Synchronous multiple oral squamous cell carcinoma in a female patient: A case report and review

Debasish Pramanick, Sandip Ghose

From Department of Oral Pathology, Dr. R Ahmed Dental College and Hospital, Kolkata, West Bengal, India

Correspondence to: Debasish Pramanick, Dr. R Ahmed Dental College and Hospital, 114, A.J.C. Bose Road, Kolkata - 700014. West Bengal, India. Phone: +91-9775928870. E-mail: dr.pramanick.debasish@gmail.com

Received - 16 September 2017 Initial Review - 12 October 2017 Published Online -30 November 2017

ABSTRACT

Oral squamous cell carcinoma (OSCC) is a common oncological problem in India. However, the frequency of developing synchronous carcinomas, i.e., development of second primary tumor (SPT) either simultaneously or within 6 months of the index tumor, in the orofacial region are rare and ranges from 8% to 21%. These lesions are more aggressive, treatment-resistant, and metastasize early. The onset of SPT decreases the 5-year survival by 18–30% as compared to those with a single tumor. Astonishingly, it was found that synchronous OSCCs showed 100% male predominance, with no female predilection. This case of synchronous OSCC in a 58-year-old female had an index tumor involving right side of the mandible and a SPT in the left buccal mucosa. Both the lesions were histopathologically diagnosed well-differentiated squamous cell carcinoma. Hence, the importance of reporting this case lies on the rarity, aggressiveness, and poor prognosis of the lesion and the patient being a female.

Key words: *Oral squamous cell carcinoma, Synchronous, Tobacco*

Oral squamous cell carcinoma (OSCC) is a carcinoma with squamous differentiation arising from the mucosal epithelium [1]. It is most frequent in the fifth and sixth decades of life and is typically associated with risk factors such as smoking, alcohol consumption, and betel-quid Chewing [1]. More than 90% of cancers encountered in the oral cavity are OSCCs. High incidence of oral cancer is found in southern Asian countries like India. Oral cancer can affect any area of the oral mucosa. Patients usually present with a single lesion involving a small area or a large lesion extending into various parts of the oral mucosa. However, the frequency of developing synchronous carcinomas in the head and neck mucosal sites are not very common and ranges from 8% to 21%. Synchronous carcinomas are those lesions where a second primary tumor (SPT) develops either simultaneously or within 6 months of the index tumor. This type of lesion is more aggressive as well as treatment resistant, and on the other hand metastasizes early, thereby requiring a more extensive treatment strategy. Moreover, according to various series, the onset of SPT decreases the 5-year survival by 18–30% as compared to those with only a single tumor [2]. Besides this, various studies on synchronous OSCCs showed 100% male predominance, with no predilection to female patients [3]. Hence, the importance of reporting this case lies on the rarity, aggressiveness, and poor prognosis of the lesion and the patient being a female.

CASE REPORT

A 58-year-old female patient, housewife came to the Department of Oral and Maxillofacial Pathology Outpatient clinic in March 2017 with the complaint of intense pain and ulceration associated to both sides of the oral cavity for the duration of 6 months. According to her report, there had been a reddish ulcerative growth in oral cavity bilaterally (Figs. 1 and 2). Ulceroproliferative growth first appeared on the right side of lower jaw 6 months back. 2 months after the first growth, a second ulceroproliferative growth evolved in the left cheek mucosa. Both the lesions grew rapidly and were refractory to any kind of medicines that she had taken after consultation with some local doctors. On physical examination, two separate lesions with extensive ulceration, surface granularity, rolled margins, and indurated borders were noted in alveolobuccal complex of the right side of the mandible and retrocommissural buccal mucosa of the left side. Lesion on the right side measured approximately about 3 cm × 2 cm while that of the left side was 2 cm × 1 cm. Midline was totally free from any abnormality, and two lesions were separated from each other by 4 cm (approx.). Bilateral submandibular lymph nodes were detected on palpation. Nodes were firm, non-tender and mobile in nature. Medical history of the patient had no important episodes. Patient admitted about the habit of chewing pan masala as well as betel quid with tobacco 3 to 4 times a day since past 4 to 5 years. Her family history was nothing significant. When hematological investigations such as routine blood, coagulation profile, glycemic status, and pre-operative serology were found to be within normal limits, incisional biopsy from representative sites was performed from two separate lesions. Histopathological examination of two separate lesional tissue sections (one from alveolobuccal complex of the right side of

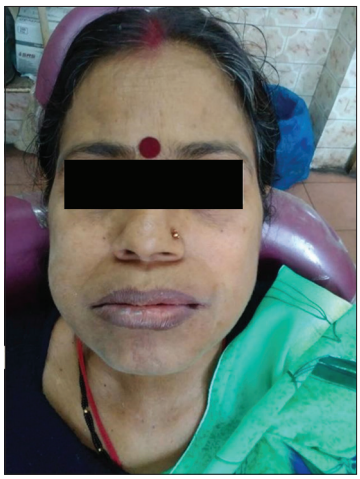

Figure 1: Frontal profile of the patient

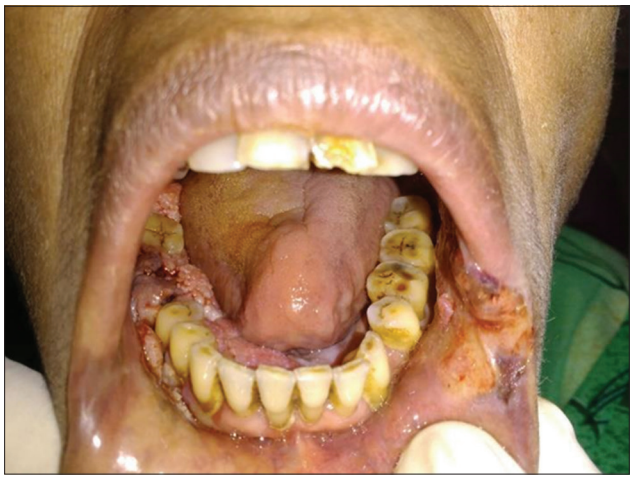

Figure 2: Intraoral view showing two separate lesion

mandible and the other from retrocommissural buccal mucosa of left side) stained with H and E revealed neoplastic epithelial islands invading into the underlying connective tissue stroma. Epithelial cells showed dysplastic features such as cellular pleomorphism, nuclear pleomorphism and hyperchromatism, and mitotic figures. Abundant keratin pearl formation was also noted. Inflammatory cell infiltrate within the connective tissue stroma was present. Overall, histopathological features were corroborative to well differentiated squamous cell carcinoma both in alveolobuccal complex of the right side (Fig. 3) of the mandible and retrocommissural buccal mucosa of the left side (Fig. 4). The patient was immediately referred to the Department of Oral Surgery for further management.

DISCUSSION

Synchronous OSCC is one of the most challenging lesions encountered in oral pathology as well as oral and maxillofacial surgery practice. Understanding the pathophysiology of it is often difficult. To account for the development of multiple (multicentric) primary tumors in the oral cavity, the concept of field cancerization by Slaughter *et al*. [4] can be interpreted in various ways to explain this phenomenon of SPTs.

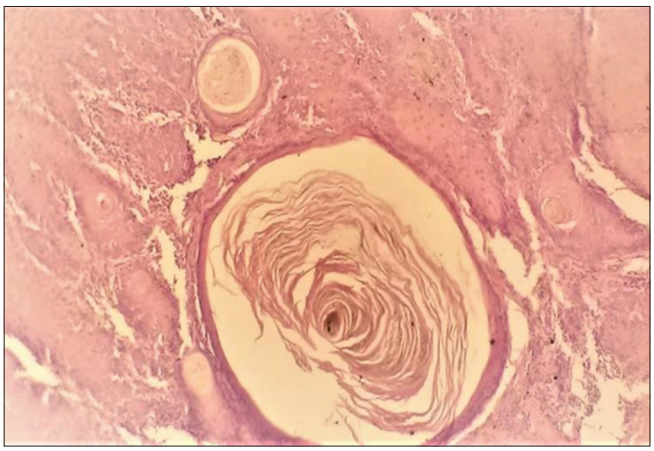

Figure 3: Histopathology of right-sided malignant ulcer showing neoplastic epithelial islands within connective tissue stroma and keratin pearl formation

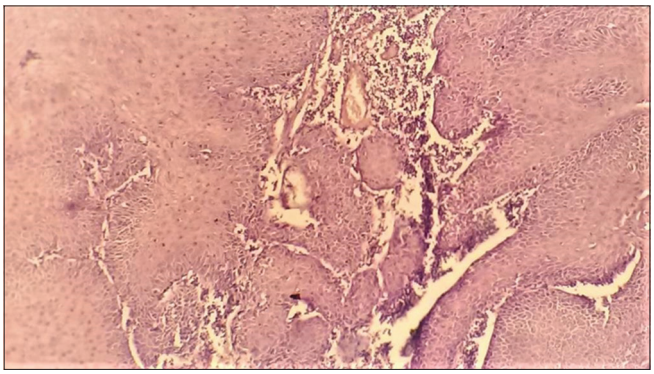

Figure 4: Histopathology of left-sided malignant ulcer showing neoplastic epithelial islands within connective tissue stroma

In the "classical view," which is most commonly referred to large areas of the aerodigestive tissue are affected by long-term exposure to carcinogens. In this preconditioned epithelium, multifocal carcinomas can develop as a result of independent mutations and thus would not be genetically related [5]. In "alternative view" also known "clonal theory," a single cell is transformed and gives rise to one large extended premalignant field by clonal expansion and gradual replacement of normal mucosa. In this field of various subclones, two separate tumors can develop after the accumulation of additional genetic alterations. Both tumors have the same clonal origin would thus share at least one early genetic event, which occurred before the initial clonal expansion [6]. Now to establish a case to be synchronous lesion, and for the diagnosis of SPT, following are the criteria suggested by Warren and Gates [7] and Moertel *et al*. [8].

1. All the neoplasms had to be histologically malignant.
2. All had to be discrete masses separated by normal tissue (at least by 2 cm) (if the intervening mucosa demonstrates dysplasia, it is considered as multicentric primary).
3. The possibility that the tumors could be metastatic had to be histologically excluded.
4. Secondary lesions had to be solitary and histologically distinct from the primary tumor.

The patient discussed in this case met all the above criteria and was diagnosed to be synchronous multiple OSCC. Aggressive treatment strategy and post-treatment regular follow-up are indispensable for such patients.

CONCLUSION

Synchronous OSCC being highly aggressive and having a poor prognosis, early diagnosis is very much important. Even though after extensive surgical management and radiotherapy, there is a high chance of locoregional recurrence.

REFERENCES

1. El-Naggar AK, Chan JK, Grandis JR, Takata T, Slootweg PJ. WHO Classification of Head and Neck Tumors. 4th ed. Lyon: International Agency for Research on Cancer (IARC); 2017. p. 109.
2. Cianfriglia F, Di Gregorio DA, Manieri A. Multiple primary tumours in patients with oral squamous cell carcinoma. Oral Oncol 1999;35:157-63.
3. Dissanayaka WL, Jayasooriya PR, Kumarasiri PV, Tilakaratne WM. A histopathologic comparison between synchronous and single primary oral squamous cell carcinomas. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2010;109:732-8.
4. Slaughter DP, Southwick HW, Smejkal W. Field cancerization in oral stratified squamous epithelium; clinical implications of multicentric origin. Cancer 1953;6:963-8.
5. Chung KY, Mukhopadhyay T, Kim J, Casson A, Ro JY, Goepfert H, *et al.* Discordant p53 gene mutations in primary head and neck cancers and corresponding second primary cancers of the upper aerodigestive tract. Cancer Res 1993;53:1676-83.
6. Jayam R. Oral field cancerization: A review. J Indian Acad Oral Med Radiol 2010;22:201-5.
7. Warren S, Gates O. Multiple primary malignant tumors: Survey of the literature and statistical study. Am J Cancer 1932;16:1358-414.
8. Moertel CG, Dockerty MB, Baggenstoss AH. Multiple primary malignant neoplasms. I. Introduction and presentation of data. Cancer 1961;14:221-30.

Funding: None; Conflict of Interest: None Stated.

How to cite this article: Pramanick D, Ghose S. Synchronous multiple oral squamous cell carcinoma in a female patient: A case report and review. Indian J Case Reports. 2018;4(1):28-30.