CASE REPORT

Second primary oral melanoma: A rare presentation

Meghanand T Nayak, Anjali Singh, Mathur RM, Puneet Wadhwani
Department of Oral and Maxillofacial Pathology, Saraswati Dental College and Hospital, Lucknow, India

Address for correspondence:
Dr. Meghanand T Nayak,
Vyas Dental College and Hospital, KudiHoud,
Pali Road, Jodhpur, Rajasthan, India.
E-mail: drmeghanand@gmail.com

ABSTRACT

Melanomas are neoplasms of melanocytic origin. They are aggressive neoplasms with an unpredictable behavior, and can involve virtually any organ of the body. Oral melanomas are very rare and have an extremely poor prognosis. Early diagnosis and prompt treatment is the key to reduce the morbidity and mortality. A second primary tumor is a new primary tumor developing in a person with a history of tumor, in a new site or tissue and subsequent to the initial tumor. Patients with previous history of melanoma are associated with a higher risk of developing second primaries. A case of second primary oral melanoma in a 55-year-old female is reported here. The anachronistic presentation of the primary and the second primary lesions make this case clinically interesting. Noteworthy immunohistochemical findings were recorded, HMB-45 positive and S-100 negative.

Key words: Buccal mucosa, follow-up, HMB-45, hard palate, oral melanoma, second primary malignancies

INTRODUCTION

Melanoma is a malignant neoplasm of the melanocytes, which are derived from neural crest cells. Although most melanomas arise from the skin, they may also arise from mucosal surfaces or other sites where neural crest cells migrate.[1]

Primary mucosal melanomas are exceptionally rare and accounts for only 0.2–8% of all melanomas.[2] Clinically, metastatic melanoma involving the skin appears as a dermal nodule or plaque-like lesion, often in close proximity to the primary tumor.[3] The development of second primary neoplasms in patients with cutaneous melanomas has been previously reported. Here, we are reporting first case of second primary oral melanoma.

CASE REPORT

A 55-year-old female reported to our institution complaining of a blackish discoloration on the left cheek since 5–6 months. Patient also complained of a feeling of discomfort during normal mastication in the region. The patient revealed that she was previously diagnosed as having melanoma on the anterior part of the hard palate and she had undergone partial maxillectomy followed by radiotherapy 7 years back.

Her previous clinical records revealed that the earlier lesion was blackish macular discoloration on hard palate, palatal to permanent left maxillary central incisor (FDI # 21) anteriorly and extending to the level of permanent maxillary left second premolar (FDI # 25) posteriorly. The mesial margin was encroaching upon the midline, while the distal margin was irregular and about 1–2 cm away from the alveolar ridge. Partial maxillectomy including both halves of maxilla was performed to remove the primary growth and an adequate free margin of 4 cm was included. The patient had uneventful recovery. The surgical defect was covered with an obturator. Her excisional biopsy confirmed the diagnosis of malignant melanoma (Clark's Level II). The patient was later treated with chemotherapy and radiotherapy. Postoperative radiotherapy records revealed that radiation was given by Cobalt-60-4400 cGy on 22 Fc. to whole neck for 20 days. A total dose of 6000 cGy in two phases was advised; however, the treatment was withdrawn after 22 Fc. as the patient had symptoms of radiation mucositis.

Physical examination of the present lesion revealed a solitary, exophytic, nodular growth measuring about 3 × 2 × 0.5 cm on left posterior buccal mucosa in relation to permanent mandibular left III molar (FDI # 38). The lesion extended from buccal aspect of mandibular III molar anteriorly to 0.5 cm posteriorly over retromolar region. The lesion was well defined and elevated, with nodular surface. The color of the lesion was variegated [Figure 1]. No other lesions were found elsewhere

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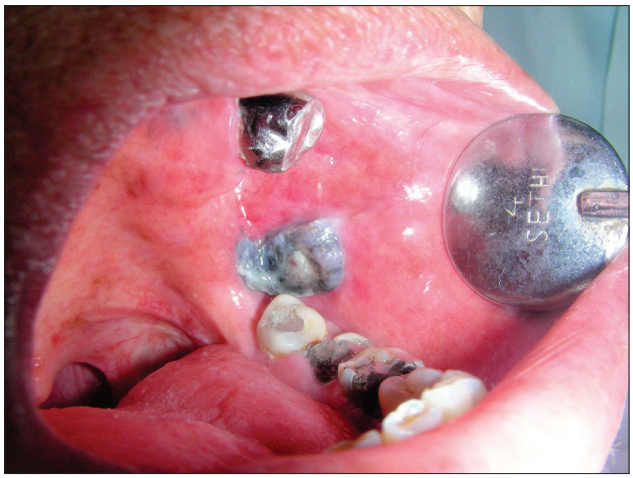

Figure 1: Intraoral photograph showing the nodular and variegated appearance of the tumor

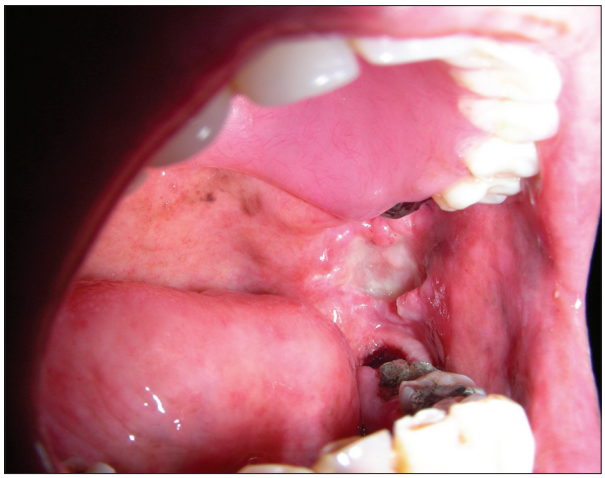

Figure 2: Tumor bed after complete excision of the tumor mass

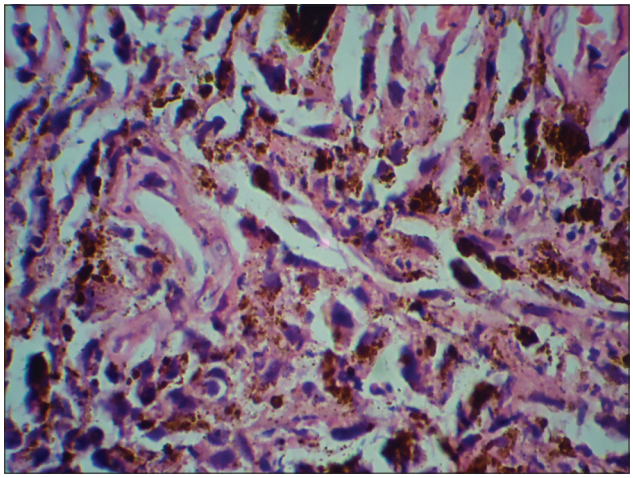

Figure 3: Histopathology reveals tumor mass composed of proliferating ovoid or polygonal cells with hyperchromatic nuclei and melanin pigmentation (H and E, stain ×40)

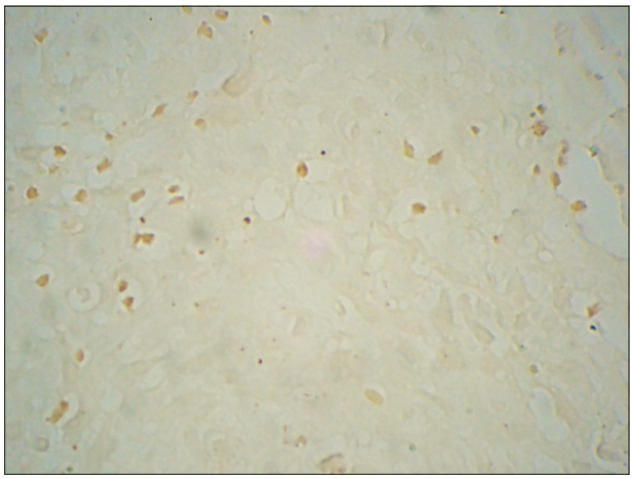

Figure 5: Immunohistochemical analysis of S-100 showing focal positivity or negative staining (×40)

in the body and no lymph nodes were palpable. The lesion was excised completely and was subjected to biopsy [Figure 2].

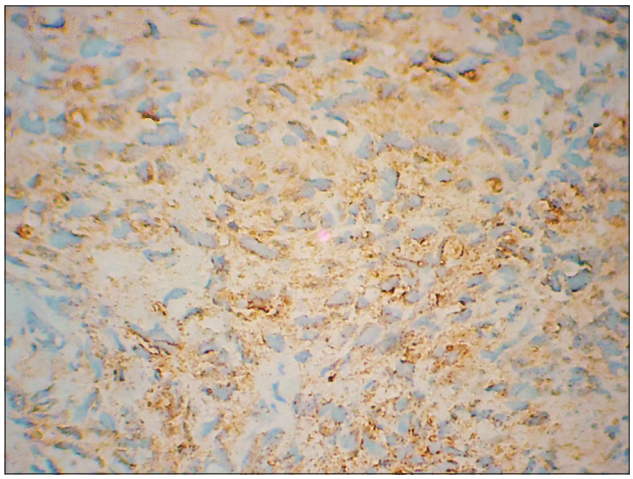

Figure 4: Immunohistochemical analysis of HMB-45 showing strong positivity, hence revealing melanocytic origin (×40)

The sections of excisional biopsy revealed fibrous connective tissue stroma infiltrated by islands of tumor mass. The tumor was composed of proliferating ovoid or polygonal cells, which were disposed diffusely. The cells exhibited hyperchromatic round nuclei and showed moderate anisocytosis [Figure 3]. Melanin pigmentation was seen in most part of the tumor. A diagnosis of "Nodular melanoma with Clark's level IV involvement" was made. A combined radiotherapy and immunotherapy was implemented to the patient and she has now successfully survived for over 10 years from the initial onset of the disease. Patient underwent radiotherapy and immunotherapy and has survived for 10 years.

DISCUSSION

Oral melanoma is a rare entity occurring at a frequency of less than 2% of the all the head and neck cancers.[4] Men are affected about 3.5 times more commonly than the women. The ethnic groups most commonly affected by oral melanomas are Japanese, black Africans, American Indians, and Hispanics.[5] This malignancy is more frequently seen on

the hard palate and maxillary gingiva.[6] Other oral sites are mandible, tongue, buccal mucosa, and upper and lower lip.

When the melanocytes or their precursor cells turn neoplastic, they possibly lead to malignancies which usually metastasize to adjacent tissues. The rich vascular supply present in the oral cavity contributes to the dissemination of the melanomas.[7]

A thorough clinical examination of any pigmented lesion that may exhibit growth potential must be done and biopsied to rule out melanomas. Melanomas metastasize first to the regional lymph nodes and then to secondary sites, most commonly skin, subcutaneous soft tissue, lung, and brain. In primary melanomas (stage I and II) recurrences are local-regional, while in patients with stage III disease recurrences are usually systemic. In our case, the lymph nodes were not palpable and the aspirations were negative; we could not find any metastatic deposits.

A second primary melanoma is a new or de novo primary melanoma that arises at the periphery of a previously excised melanoma, in the context of a field defect. It is important to differentiate an additional melanoma from a metastatic melanoma, because the prognosis of an independent primary lesion is likely to be much better than that of a metastasis.[8]

Increased incidence of germ-line mutations has been noted in CDKN2A locus in patients with multiple primary melanomas.[9,10] In this case, the first primary lesion; occurred on the hard palate was superficial and indolent, while the second primary lesion which occurred after a gap of 7 years was present on the buccal mucosa and was nodular in nature with variegated surface. Probably this change in presentation was in response to the stepwise accumulation of genetic abnormalities. Nodular melanomas are vertical growth phase melanomas and comprise 15% to 30% of all melanomas.[11] They usually appear as a darkly pigmented, pedunculated, or polypoid nodule. Our case had a similar presentation.

The antibody HMB-45 and S-100 reacts with a 10-kDa cytoplasmic glycoprotein that is part of the premelanosome complex.[12,13] A positive HMB-45 immunostaining and a focal or absent S-100 staining was noted in our case [Figures 4 and 5]. It has been previously reported that in a small subset of melanomas S-100 protein is either not expressed or is expressed at a level below that which can be detected by routine immunohistochemistry.[14] Nevertheless, a positive HMB-45 reactivity provides a strong evidence of melanocytic histiogenesis.

For individuals with a history of a prior cutaneous melanoma, the risk of a second primary is very high.[14] Although the risk is highest in the first year after initial diagnosis, the risk of developing a second melanoma have ranged from 2–11% at 5 years.[15,16]

The most important elements in the follow-up of oral melanoma patients are medical history and physical examination, paying particular attention to recurrences. To the best of our knowledge this report documents the first case of second primary oral melanoma occurring in a previously treated patient, who has survived over 10 years.

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