

## Primary Malignant Melanoma of Oral Mucosa – Report of Two Cases

### Abstract

Primary malignant melanoma of oral mucosa is a rare and aggressive tumor. It is usually seen in the 5<sup>th</sup> and 6<sup>th</sup> decades of life. Its mainstay of treatment is surgery. It has a very poor prognosis, which is attributed to its late detection and distant metastasis. Dentists are often the first clinicians to come across these lesions and need to be able to identify them at the earliest for a better prognosis. In this article, we present two cases of extensive primary malignant melanoma of the oral cavity. Clinically, both the cases had a similar appearance of grayish-black pigmented nodular swelling on the buccal aspect and grayish-black discoloration on the palatal aspect. There were no significant radiological changes in both cases, indicating the superficial spread of the lesion. A positron emission tomography scan was performed in the second patient, which did not show any distant metastasis. Surgery was advised as a treatment for both the patients.

**Keywords:** *Gingiva, hard palate, oral mucosa, primary malignant melanoma*

### Introduction

Primary malignant melanoma of the oral cavity is a rare tumor arising from melanocytes or melanocytic precursors.<sup>[1-3]</sup> Melanocytes are neural crest-derived cells that migrate to the skin, mucous membrane, and several other sites. In the skin, they protect from ultraviolet radiation and sun exposure. The function of melanocytes in the mucosa is not fully understood.<sup>[4]</sup> Malignant melanoma comprises 3%–5% of all cutaneous malignancies and its oral cavity counterpart accounting for 0.2%–8% of all malignant melanomas.<sup>[5]</sup> The first case of primary oral malignant melanoma was reported by Weber in 1859.<sup>[1]</sup> Oral malignant melanomas have a slightly higher incidence in countries such as Japan, Uganda, and India.<sup>[6-8]</sup> It is usually seen in the 5<sup>th</sup> and 6<sup>th</sup> decades of life, and males are twice more commonly affected than females.<sup>[8-10]</sup> Etiology of oral malignant melanomas are unknown, although it has been frequently linked to excessive pigmentation of oral mucosa and benign melanotic oral lesions.<sup>[3]</sup> Oral mucosal melanoma is derived from melanocytes present in the ectodermally derived mucosa with neuroectodermal origin now classified as a depressed neuroendocrine cell system, which may explain its low incidence in

the mucosa of endodermal origin.<sup>[7]</sup> In the oral cavity, the most common site of origin is the hard palate, followed by buccal gingiva.<sup>[8,9]</sup> Clinically, oral malignant melanomas are often asymptomatic and can present as melanotic or nonmelanotic lesions. The color ranges from black to gray to purple to red to white, and lesions appear asymmetric, irregular in outline, and occasionally multiple.<sup>[9]</sup> The melanotic form may present as nodular, macular, or mixed type.<sup>[2]</sup> The management of oral malignant melanomas requires early and aggressive intervention.<sup>[9]</sup> Surgery is the mainstay of treatment. Although radiotherapy, chemotherapy, and immunotherapy have been used, they have demonstrated a relatively low response rate.<sup>[8]</sup> The prognosis of oral malignant melanoma is very poor. The 5-year survival rate has been reported to be 15%, with a median survival of 25 months.<sup>[4]</sup> One of the factors attributed to the poor prognosis is the lack of data about clinical behavior due to the rarity of this condition.<sup>[8,11]</sup> Dentists are often the first to come across oral malignant melanomas. Dentists need to be able to recognize oral malignant melanoma to minimize any delay in the treatment and contribute to the better prognosis of the condition. In this regard, we present two cases of primary oral malignant melanoma extensively involving the buccal gingiva and hard palate region.

This is an open access journal, and articles are distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 4.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as appropriate credit is given and the new creations are licensed under the identical terms.

For reprints contact: reprints@medknow.com

**How to cite this article:** Chandan SN, Shetty SK, Deepa BV. Primary malignant melanoma of oral mucosa – Report of two cases. *Contemp Clin Dent* 2020;11:195-8.

**S. N. Chandan,  
Sahith Kumar  
Shetty, B. V. Deepa**

*Department of Oral and Maxillofacial Surgery, JSS Dental College and Hospital, JSS Academy of Higher Education and Research, Mysuru, Karnataka, India*

Submitted : 05-Nov-2019

Revised : 09-Nov-2020

Accepted : 17-Apr-2020

Published : 07-Aug-2020

### Address for correspondence:

*Dr. S. N. Chandan,  
Department of Oral and Maxillofacial Surgery, JSS Dental College and Hospital, JSS Academy of Higher Education and Research, Mysuru - 570 015, Karnataka, India.  
E-mail: drchandansn@gmail.com*

### Access this article online

#### Website:

[www.contempclindent.org](http://www.contempclindent.org)

DOI: 10.4103/ccd.ccd\_380\_19

#### Quick Response Code:



## Case Reports

### Case 1

A 40-year-old male patient visited a dentist with a complaint of nonhealing growth over the labial gingiva. The patient first noticed a small painless swelling in the upper anterior gingival region 3 months back. The dentist provisionally diagnosed it as pyogenic granuloma and treated with oral prophylaxis, antiseptic mouth wash. The patient again visited the dentist 3 months later, at which time the lesion had enlarged significantly and was referred to the authors. The patient was a farmer by occupation and had no relevant medical or family history. He was a betel nut chewer, smoker with 2–3 beedis per day and did not consume alcohol. Intraorally, a nontender, grayish-black nodular swelling was seen extending from the second premolar on one side to the other on labial gingiva slightly extending into the vestibule [Figure 1a]. A grayish-black discoloration of palatal mucosa was seen [Figure 1b]. Clinically, there were no palpable cervical lymph nodes. An orthopantomogram and maxillary occlusal radiograph were taken. Both appeared normal and did not indicate any erosion or bony invasion of the tumor. An incision biopsy was performed, and histopathological report diagnosed it as malignant melanoma. The surgical excision of the lesion was planned after a positron emission tomography (PET) scan to identify any metastasis. The prognosis of the lesion was explained to the patient. The patient was lost for follow-up.

### Case 2

A 50-year-old male patient came with the complaint of painless growth in the upper gingiva for 6 months. The patient was a cab driver by occupation. The patient had no relevant medical or family history. The patient was a chronic smoker with 30 cigarettes per day for 25 years and consumed alcohol occasionally. Intraorally, on the left side, a nontender, grayish-black nodular swelling noted

extending from midline to first molar region on maxillary labial gingiva. A grayish-black discoloration of labial attached gingiva was seen on the right side extending from midline to canine region [Figure 2a]. On the palatal side, grayish-black discoloration was noted, extending from the lateral incisor region on the right side to the left side third molar region [Figure 2b]. A 1 cm × 1 cm soft, nontender, mobile lymph node was palpable in the left submandibular region. An orthopantomogram and maxillary occlusal radiograph were taken. Both appeared normal and did not indicate any erosion or bony invasion of the tumor. An incision biopsy was performed and was diagnosed as malignant melanoma [Figure 3]. A PET scan was performed, which did not show any distant metastasis. The patient was explained of nature of the surgery required and the prognosis of the lesion. The patient wanted treatment in a different city and was referred for further management.

## Discussion

Oral malignant melanoma is a rare and aggressive condition. Due to their asymptomatic nature, they are often diagnosed at a much later stage of the disease. This, in combination with a lack of information on the etiology and unreliable clinical detection methods, makes this tumor a very difficult entity to manage.<sup>[12]</sup> In the above cases, the patients were both males and were in their 5<sup>th</sup> decade of life, which is consistent with the presentation of oral melanomas. Oral melanomas commonly occur in maxillary gingiva, hard palate, and alveolar ridge, and only 20% of these lesions are observed in the mandibular mucosa.<sup>[3]</sup> Clinically both the above cases were melanotic type and had a similar appearance of grayish-black pigmented nodular swelling on the buccal aspect and grayish-black discoloration on the palatal aspect. It is important to note that 2% to 40% of oral malignant melanomas are amelanotic with an absence of pigmentation and have a significantly poorer prognosis.<sup>[13]</sup> Melanomas are known to spread superficially and only involve deeper tissues at a later stage. This

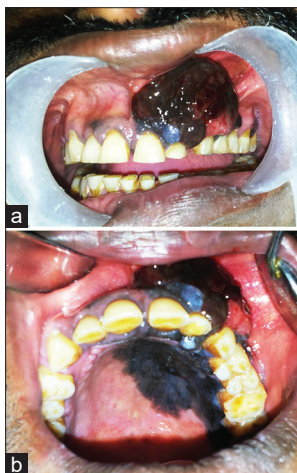


Figure 1: (a) Grayish-black nodular swelling and discoloration of labial attached gingiva. (b) Grayish-black discoloration of palatal mucosa

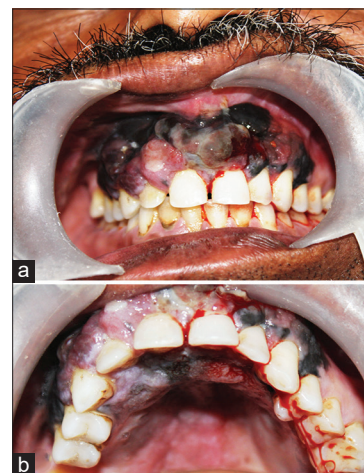
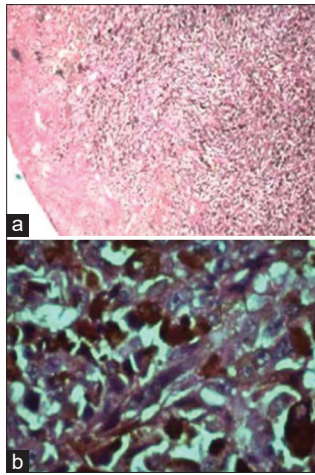


Figure 2: (a) Grayish-black nodular swelling on labial mucosa. (b) Grayish-black discoloration of palatal mucosa



**Figure 3:** (a) Connective tissue stroma with sheets of tumor cells containing melanin pigment (H and E,  $\times 100$ ). (b) Atypical melanocytes (H and E,  $\times 400$ )

explains their hematological distant metastasis rather than lymphatic. During the incision biopsy of the above cases, it was noted in both cases the underlying periosteum and bone appeared to be normal, indicating the lateral spread of the tumor. In both patients, the tumor was painless, hence the neglect and late presentation. Dentists usually are the first clinicians to notice these lesions and play an important role in the diagnoses of oral malignant melanoma. During any routine oral examination, one should make a note of any pigmented lesions and have a low threshold to biopsy any suspicious lesions.

Most oral melanomas are large at presentation and have a poorer prognosis than cutaneous melanomas.<sup>[8]</sup> Cutaneous melanomas can be graded by Clark levels or the Breslow's tumor thickness grading system. The former classification assesses the depth of invasion, whereas Breslow's system measures the thickness of the tumor from the surface of the epidermis to the greatest depth of the tumor. The risk for developing metastatic lesions from primary cutaneous melanomas increases with tumor thickness. The Breslow and Clark grading systems have not been validated as prognostic predictors in oral melanomas, probably owing to the rarity of this lesion. In addition, in contrast to cutaneous melanomas, most oral melanomas are larger than 4 mm at the time of the initial presentation. With histopathological examination, oral melanomas can be easily diagnosed on H and E, stained sections and when the pigment is completely absent, we can use immunohistochemical markers like S-100, gp100 (HMB-45), and Mart-1 (Melan-A). Even tumor node metastasis (TNM) staging for cutaneous melanomas needs to be applied with caution while treating oral melanomas.<sup>[14]</sup> According to the American Joint Committee on Cancer (AJCC) TNM classification for mucosal melanoma of the head and neck,<sup>[15]</sup> both the above cases were T3, N0, M0, making them be in Stage III of the disease. It is to be noted that the AJCC staging for mucosal melanomas of the head and neck begins at Stage III owing

to the extensive tumor during the initial presentation. This has tremendous implications for prognosis. Together with inadequate resection of margins and higher stage at initial diagnosis, may contribute to the discrepancy in patients' 5-year survival rates between cutaneous melanoma (80%) and oral melanomas (15%).<sup>[4]</sup> In the above cases, there was no distant metastasis in the second case as determined by the PET scan, whereas it could not be determined in the first case. Surgical resection remains the treatment of choice for oral malignant melanomas. Although previously melanoma was considered to be radioresistant, radiotherapy is now considered to be an important adjuvant treatment for local control.<sup>[11]</sup> Other adjuvant therapies such as immunotherapy and chemotherapy are also advocated and are showing promising results. Surgery was advised as a treatment modality in both the cases and the prognosis was explained. One case was lost for follow-up, and the other was referred to a cancer center for further management.

#### 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.

#### Financial support and sponsorship

Nil.

#### Conflicts of interest

There are no conflicts of interest.

#### References

1. Chaudhry AP, Hampel A, Gorlin RJ. Primary malignant melanoma of the oral cavity: A review of 105 cases. *Cancer* 1958;11:923-8.
2. Manigandan T, Sagar GV, Amudhan A, Hemalatha VT, Babu NA. Oral malignant melanoma: A case report with review of literature. *Contemp Clin Dent* 2014;5:415-8.
3. Deyhimi P, Razavi SM, Shahnasari S, Khalesi S, Homayoni S, Tavakoli P. Rare and extensive malignant melanoma of the oral cavity: Report of two cases. *J Dent (Shiraz, Iran)* 2017;18:227-33.
4. Hicks MJ, Flaitz CM. Oral mucosal melanoma: Epidemiology and pathobiology. *Oral Oncol* 2000;36:152-69.
5. Boring CC, Squires TS, Tong T, Montgomery S. Cancer statistics. *CA Cancer J Clin* 1994;44:7-26.
6. Takagi M, Ishikawa G, Mori W. Primary malignant melanoma of the oral cavity in Japan. With special reference to mucosal melanosis. *Cancer* 1974;34:358.
7. Pandey M, Mathew A, Iype EM, Sebastian P, Abraham EK, Nair KM. Primary malignant mucosal melanoma of the head and neck region: Pooled analysis of 60 published cases from India and review of literature. *Eur J Cancer Prev* 2002;11:3.
8. Hashemi Pour MS. Malignant melanoma of the oral cavity: A review of literature. *Indian J Dent Res* 2008;19:47-51.
9. Barker BF, Carpenter WM, Daniels TE, Kahn MA, Leider AS,

- Lozada-Nur F, *et al.* Oral mucosal melanomas: The WESTOP Banff workshop proceedings. Western Society of Teachers of Oral Pathology. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1997;83:672-9.
10. Wang X, Wu HM, Ren GX, Tang J, Guo W. Primary oral mucosal melanoma: Advocate a wait-and-see policy in the clinically N0 patient. J Oral Maxillofac Surg 2012;70:1192-8.
  11. Lamichhane NS, An J, Liu Q, Zhang W. Primary malignant mucosal melanoma of the upper lip: A case report and review of the literature. BMC Res Notes 2015;8:499.
  12. Smith MH, Bhattacharyya I, Cohen DM, Islam NM, Fitzpatrick SG, Montague LJ, *et al.* Melanoma of the oral cavity: An analysis of 46 new cases with emphasis on clinical and histopathologic characteristics. Head Neck Pathol 2016;10:298-305.
  13. Feller L, Khammissa RA, Lemmer J. A review of the Aetiopathogenesis and clinical and Histopathological features of oral mucosal melanoma. ScientificWorldJournal 2017;2017:9189812.
  14. Munde A, Juvekar MV, Karle RR, Wankhede P. Malignant melanoma of the oral cavity: Report of two cases. Contemp Clin Dent 2014;5:227-230.
  15. Head and Neck. American Joint Committee on Cancer. AJCC Cancer Staging Manual. 8<sup>th</sup> ed. New York, NY: Springer; 2016.