

Primary retromolar malignant melanoma in a Latin American patient

Journal of International Medical Research 48(4) 1–6 © The Author(s) 2020 Article reuse guidelines: sagepub.com/journals-permissions DOI: 10.1177/0300060520919245 journals.sagepub.com/home/imr



Carlos-Martín Ardila[®], Joan-Michael Arce-Jaramillo and Efraín Álvarez-Martínez

Abstract

Primary malignant melanoma of the oral mucosa is rare and has scarcely been reported in patients from Latin America. The most frequently affected sites include the palate and gingiva of the maxilla. In this report, we describe primary malignant melanoma in a Latin American patient. A 3×3.5 -cm black tumor was observed on the upper left retromolar area. The lesion exhibited a sessile base, irregular edges, soft consistency, and pain on palpation. Immunohistochemistry staining results were positive for S100 and HMB-45; the Ki-67 index was 20%. Contrast tomography of the skull, face, abdomen, and thorax, as well as ultrasound of the liver, did not reveal metastatic lesions. The patient underwent wide surgical excision of the lesion, followed by radiotherapy; he was subsequently rehabilitated with a somato prosthesis.

Keywords

HMB-45 protein, immunohistochemistry, melanoma, malignant, melanocytes, Latin America, oral mucosa

Date received: 30 September 2019; accepted: 5 March 2020

Introduction

Malignant melanoma of the oral mucosa is both rare and highly lethal.^{1–3} The etiology of this neoplasia is unknown, although it may arise from a preexisting benign melanocytic lesion or nevus.^{4,5} It is painless in its early stages, with asymmetric and irregular contour.⁶ Diagnosis is often delayed, and is

Universidad de Antioquia, Medellín, Colombia

Corresponding author: Carlos Martín Ardila, Universidad de Antioquia, Calle 70 No. 52-21, Medellín, Colombia. Email: martin.ardila@udea.edu.co

Creative Commons Non Commercial CC BY-NC: This article is distributed under the terms of the Creative Commons Attribution-NonCommercial 4.0 License (https://creativecommons.org/licenses/by-nc/4.0/) which permits non-commercial use, reproduction and distribution of the work without further permission provided the original work is attributed as specified on the SAGE and Open Access pages (https://us.sagepub.com/en-us/nam/open-access-at-sage).

only made when symptoms manifest as a result of ulceration or growths. The melanoma becomes a strongly pigmented tumor, sometimes ulcerated and hemorrhagic, which tends to increase in size.^{4,7–9} The most commonly affected mucous membranes include the conjunctiva, upper respiratory tract, nasal cavity, paranasal sinuses, and oral cavity. Within the oral cavity, the most frequently affected sites include the palate and maxillary gingiva; combined, these sites exhibit 80% of all melanomas in the oral cavity.^{4,6,10–13} The remaining melanomas are found in the mandibular gum, oral mucosa, tongue, and floor of the mouth.⁵

Primary melanoma of the oral mucosa occurs frequently among individuals of Asian and African ethnicities, partially in relation to the high frequency of melanic pigmentation in the oral mucosa.^{1–3,6,11} To the best of our knowledge, primary malignant melanoma has scarcely been reported in patients from Latin America; reports of this subtype of melanoma are important for accurate diagnosis and treatment of Latin American individuals. Here, we describe primary retromolar malignant melanoma in a Latin American patient.

Case report

Initial presentation

A 76-year-old man presented to the Stomatology and Maxillofacial Surgery service at San Vicente Hospital in Medellín, Colombia. Before any examination or intervention, the patient provided written informed consent for treatment. The patient had worked as a farmer for 60 years and had smoked approximately 10 cigarettes per day for 20 years; his smoking habit was ongoing at the time of presentation for treatment. The hospital's general dentistry service had referred the patient for treatment by the Stomatology and Maxillofacial Surgery service, due to the presence of a mass that had evident for 8 months in the left superior alveolar ridge. The patient reported that the lesion had been caused by abrasion from an encrusted fishbone; 3 months after this event, the patient experienced local edema, severe pain, and dysphagia. He also reported weight loss (20 kg in 1 year) and difficulty eating.

Intraoral examination

During intraoral examination, a 3×3.5 -cm black tumor was observed on the upper left retromolar area (Figure 1). The lesion showed a sessile base, irregular edges, rough surface, and soft consistency, as well as pain on palpation; the pain extended to the vestibular and palatal mucosa of neighboring molars. Easily bleeding mucosae were also noted with some slightly ulcerated areas. A 3-mm satellite lesion was observed in the palatal gingiva of molar #27 (Figure 1); this tooth exhibited dental mobility, but was vital. However, no translucency, compressibility, or fluctuation were observed. Left submandibular adenopathy



Figure 1. Intra-oral examination of malignant melanoma.



Figure 2. Histopathology and immunohistochemistry staining analyses of malignant melanoma lesion. (a) Stratified squamous epithelium with melanocyte infiltration (hematoxylin and eosin staining, $200 \times$); (b) positive melanin immunohistochemical staining, $100 \times$; (c) positive HMB-45 immunohistochemical staining, $100 \times$; and (d) positive S100 immunohistochemical staining, $200 \times$.

(approximately 2×2.5 cm) was evident; this was asymptomatic, with firm consistency.

Imaging and histological investigations

Further assessments included incisional biopsy; contrast tomography of the skull, face, neck, abdomen, and thorax; and liver ultrasound to establish distant metastasis. Interservice consultation with dermatology was conducted to rule out a primary origin in the skin. Biopsy revealed the presence of an acanthotic squamous epithelium with areas of ulceration and necrosis, congestive chorion, and cells occluded by black pigment in the cytoplasm, which impeded adequate evaluation; however, the presence of massive nuclei and prominent nucleoli were suggestive of a melanocytic lesion (Figure 2a). The tumor cells appeared to occupy and infiltrate deep connective tissues. Moreover, disproportionate mitotic figures were detected.

Immunohistochemistry analyses were performed to evaluate the presence of melanin, iron, S100, HMB-45, and Ki-67 in the lesion. Staining results were positive for melanin, S100, and HMB-45; the Ki-67 index was 20%. The iron staining result was negative (Figure 2b–d). These findings confirmed the diagnosis of oral melanoma. Contrast tomography of the skull, face (Figure 3a, b), abdomen, and thorax, as well as liver ultrasound, revealed normal findings without metastatic lesions; however, cervical adenopathy (3×2 cm) was observed in zone II of the neck (Figure 3c).

Outcome

Subsequently, the patient underwent wide surgical excision of the lesion (Figure 3d) and radiotherapy. He was then rehabilitated



Figure 3. Contrast tomography and surgical findings of malignant melanoma lesion. (a) Coronal contrast tomography; (b) axial contrast tomography; (c) neck contrast tomography showing an adenopathy cervical in the left area; and (d) surgical excision of the lesion.

with a somato prosthesis. Follow-up after 12 weeks of therapy revealed considerable healing at the site of intervention; there was no indication of lesion activity or remote metastases, as demonstrated by abdominal x-ray, liver ultrasound, and liver function examinations. However, 6 months later, the patient died due to metastasis of a different tumor to the lungs. Prior to his death, the patient provided consent for publication of this report.

Discussion

Malignant melanoma is an aggressive tumor with high metastatic potential; it also has the ability to invade surrounding

tissues more quickly than other oral cancers.^{3,12,13} Similar to the age of our patient, the age of onset of this melanoma ranges from 20 to 80 years (most commonly, 41-60 years); it also predominantly affects men (2:1 ratio, compared with women).^{2,5} In the oral cavity, primary malignant melanomas comprise 6.3% of all melanomas localized in the head and neck area;⁵ only approximately 0.7% to 1.6% of all melanomas originate in the oral mucosa.^{1,6,12} Although there is no ethnic predilection or environmental factor that contributes to increased incidence of this neoplasm, individuals of African and Asian ethnicity are more commonly affected than individuals of Caucasian ethnicity.^{1–3,6,11} However, considering that oral pigmented lesions have been observed frequently in Latin American mestizo patients,^{14,15} clinicians must carefully examine the oral cavity in patients with suspected cancerous lesions; some pigmented lesions should be biopsied.

As in this report, the diagnosis of oral malignant melanoma is generally confirmed by histological analysis. Possible metastasis must be verified by means of clinical examinations. The patient in this case met the criteria for primary intraoral melanoma: a clinical and microscopical demonstration of the tumor in the oral mucosa, tissue characteristics consistent with an active lesion, and the absence of any other extraoral primary site.¹⁶ Similar to the symptoms in our patient, other authors have described ulceration, hemorrhage, dental mobility, and (occasionally) pain as the most common symptoms of primary intraoral melanoma.^{1,3} Considering its clinical appearance, this melanoma was classified as a pigmented nodular type; other categories include nonpigmented nodular, pigmented macular, pigmented mixed, and non-pigmented mixed.¹⁷ Some risk factors such as tobacco use and local chronic irritations have been associated with the occurrence of oral melanoma.^{2,3,5} In the present case, the patient had smoked approximately 10 cigarettes per day for 20 years. The diagnosis of melanoma can be corroborated by positive immunohistochemistry staining results for S100, HMB-45, and Ki-67 (principally in non-melanotic melanomas);^{9,10} in biopsy from our patient, strong staining was observed for HMB-45, a preferred marker for melanocytes.¹⁰ Our patient was treated accordance with recommendations in described in the literature: wide surgical excision was performed with ample margins and adjunctive radiotherapy; chemotherapy and immunotherapy were also recommended.2,9,10

Oral mucosal melanoma is an aggressive disease with extremely poor outcome, but the 5-year overall survival rate is reportedly greater than 30%.^{18–20} Prompt diagnosis and rapid treatment of oral malignant melanoma are essential for a good prognosis.

Declaration of conflicting interest

The authors declare that there is no conflict of interest.

Funding

This research received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

ORCID iD

Carlos-Martín Ardila D https://orcid.org/0000-0002-3663-1416

References

- Markman RL, Rosa GAB, Cardili L, et al. Tongue metastasis of cutaneous melanoma: report of two cases and literature review. *J Clin Exp Dent* 2018; 10: e1130–e1134.
- Aguas SC, Quarracino MC, Lence AN, et al. Primary melanoma of the oral cavity: ten cases and review of 177 cases from literature. *Med Oral Patol Oral Cir Bucal* 2009; 14: 265–271.
- Hashemi Pour MS. Malignant melanoma of the oral cavity: a review of literature. *Indian J Dent Res* 2008; 19: 47–51.
- Gauwerky KJ, Ehrenfeld M, Bakos RM, et al. A rare case of local relapsing oral melanoma. *J Deutsch Dermatol Ges* 2010; 8: 614–616.
- Tchernev G, Lotti T and Wollina U. Palatal melanoma: "the silent killer". Open Access Maced J Med Sci 2018; 28: 364–366.
- 6. Gondivkar SM, Indurkar A, Degwekar S, et al. Primary oral malignant melanoma: a case report and review of the literature. *Quintessence Int* 2009; 40: 41–46.
- Álvarez ME, Camacho AF, Sánchez IM, et al. Melanoma of the oral mucosa with cerebral metastasis: a clinical case. Oral Oncology Extra 2005; 41: 30–33.

- Kim HS, Kim EK, Jun HJ, et al. Noncutaneous malignant melanoma: a prognostic model from a retrospective multicenter study. *BMC Cancer* 2010; 10: 167.
- 9. Rathore RS, Phulari RG, Vasavada DG, et al. A rare and extensive case of oral malignant melanoma involving mandibular gingiva. *J Clin Diagn Res* 2016; 10: ZD11–ZD12.
- Astekar M, Choubey RR, Manjunatha BS, et al. Oral malignant melanoma of alveolar ridge. *BMJ Case Rep* 2019; 12: e227456.
- Kruse AL, Riener MO, Graetz KW, et al. Mucosal malignant melanomas in head and neck surgery: a retrospective study of six patients and review of the literature. *Oral Maxillofac Surg* 2010; 14: 143–147.
- Tucci R, Aburad De Carvalhosa A, Anunciação G, et al. Late diagnosis of a primary oral malignant melanoma: a case report. *Minerva Stomatol* 2010; 59: 55–59.
- Wu AJ, Gomez J, Zhung JE, et al. Radiotherapy after surgical resection for head and neck mucosal melanoma. *Am J Clin Oncol* 2010; 33: 281–285.
- 14. Carlos-Bregni R, Contreras E, Netto AC, et al. Oral melanoacanthoma and oral melanotic macule: a report of 8 cases, review of the literature, and immunohistochemical analysis. *Med Oral Patol Oral Cir Bucal* 2007; 12: E374–E379.

- Pennacchiotti G, Oviedo C and Ortega-Pinto A. Solitary pigmented lesions in oral mucosa in Latin American children: a case series. *Pediatr Dermatol* 2018; 35: 374–377.
- Manganaro AM, Hammond HL, Dalton MJ, et al. Oral melanoma: case reports and review of the literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1995; 80: 670–676.
- Lopez-Graniel CM, Ochoa-Carrilo FJ and Menese-Garcia A. Malignant melanoma of the oral cavity: diagnosis and treatment experience in a Mexican population. *Oral Oncol* 1999; 35: 425–430.
- González-García R, Naval-Gías L, Martos PL, et al. Melanoma of the oral mucosa. Clinical cases and review of the literature. *Med Oral Patol Oral Cir Bucal* 2005; 10: 264–271.
- 19. Prasad ML, Patel S, Hoshaw-Woodard S, et al. Prognostic factors for malignant melanoma of the squamous mucosa of the head and neck. *Am J Surg Pathol* 2002; 26: 883–892.
- 20. Manigandan T, Sagar GV, Amudhan A, et al. Oral malignant melanoma: a case report with review of literature. *Contemp Clin Dent* 2014; 5: 415–418.