

MELANOMA OF THE ORAL CAVITY: A CASE REPORT

Chaidan Intapa^{1,2*}

1. D.D.S., Ph.D., Department of Oral Diagnosis, Faculty of Dentistry, Naresuan University, Phitsanulok, Thailand.

2. D.D.S., Ph.D., Department of Oncology and Diagnostic Sciences, Dental School, University of Maryland Baltimore, USA.

Abstract

The most predominant sites for oral malignant melanoma (MM) are the maxillary gingiva and hard palate that usually involve parts of the maxilla. The signs and symptoms of oral MM vary widely and clinically can present anywhere from a small irregular pigmented or discolored site, to a friable-pigmented lesion, to ulceration, to a non-pigmented mass. Many of patients do not show early symptoms, leading to most oral malignant melanomas being diagnosed at an advanced stage. The poor prognosis for this lesion relates to the amount of tumor cell vertical growth phase. However, surgery provides the best chance for long-term survival. The overall poor prognosis of oral MM must encourage health care practitioners to be careful with any changes in color or texture of oral mucosa.

Case report (J Int Dent Med Res 2014; 7: (3), pp. 56-59)

Keywords: Oral, melanoma, maxilla, maxillectomy.

Received date: 10 August 2014

Accept date: 11 November 2014

Introduction

Oral cavity is one of the most common site harboring head and neck malignant melanoma (MM).¹ Oral MM are an aggressive malignancy due to their propensity for local invasion, including into the underlying bone as well as their early metastatic spread. The most common sites of metastasis are the lymph nodes, lung, liver and bone.^{3,5,6}

The most predominant sites for oral MM are the hard palate and maxillary gingiva, which accounts for 42% and 32.1% of intraoral cases, respectively.^{2,3} Clinically, the signs and symptoms of oral MM vary widely and include anything from a small irregular pigmented or discolored site, to a large mass. Importantly, many of patients do not show early symptoms, and the average time from the manifestation of the lesion to evaluation by a health care professional is 9 months.¹ In addition, approximately one third of oral melanomas are

amelanotic leading to even more delayed diagnosis.¹ Hence, most oral MM are diagnosed at an advanced stage and usually end up with maxillectomy in most cases.

Case Report

A 50-year-old white male presented to the Oral and Maxillofacial Surgery Service at the University of Maryland, Baltimore for evaluation of the upper right maxillary dentition and possible extraction of a maxillary third molar due to a carious lesion. At the initial visit, the patient was not in any acute distress. There was no evidence of facial swelling; however, there were palpable lymph nodes at level I and II of the right neck. The patient's past medical history was significant for hypertension, for which the patient took lisinopril/hydrochlorothiazide 20/12.5mg and lisinopril 20 mg for several years. The patient reported smoking 1 pack of cigarettes per day for the past 20 years.

Upon oral examination, a 2 × 1 cm fungating, raised dark purple mass was noted which extended from the right buccal gingiva of maxillary second molar to the distal of maxillary third molar (Fig. 1A). The soft tissue lesion was friable and highly vascular. No lesions were noted on the palate or any other regions of the oral cavity.

*Corresponding author:

Dr. Chaidan Intapa
Department of Oral Diagnosis Faculty of Dentistry
Naresuan University 99 Moo9, Muang
Phitsanulok, 65000 THAILAND

E-mail: ichaidan@hotmail.com

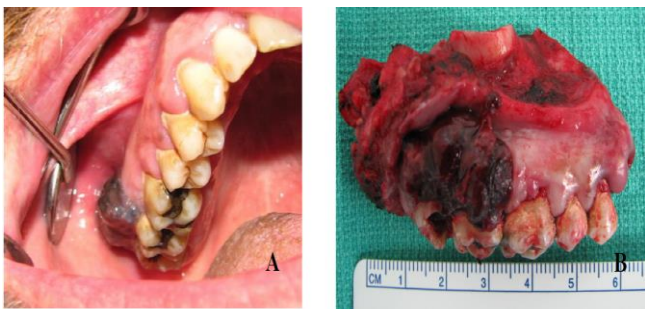


Figure 1. Photographs at initial examination showing a hyperplastic pigmented mass arising at posterior maxillary gingiva. (A) Surgical specimen at time of resection. Upper right maxilla (B)

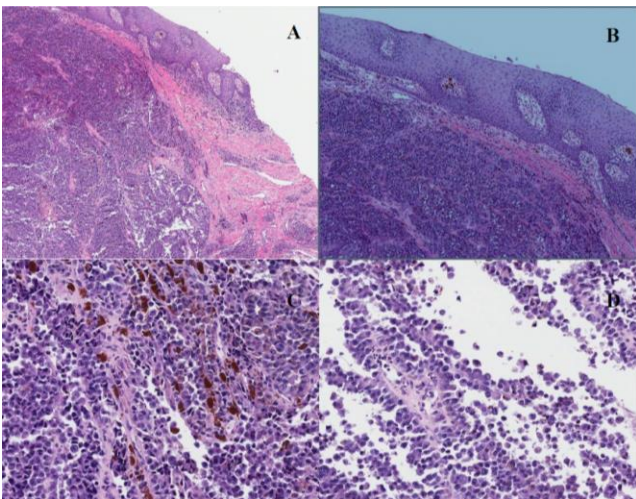


Figure 2. Tissue sections reveal infiltration of the connective tissue by neoplastic cell. (A) Groups of brown-pigmented cell were observed in connective tissue papillae as well as in neoplastic islands.(B) Tumor cells were predominantly composed of plasmacytoid cell, arranged in a solid (C) or alveolar pattern.(D) Large cells with melanin pigment deposition and neoplastic multinucleated cell were also observed. (Magnification x2) (A), (Magnification x8) (B), (Magnification x20) (C and D).

An incisional biopsy of oral mass was performed and the tissue was sent for histologic examination. Microscopic examination of the specimen demonstrated sheets of neoplastic cells surfaced by parakeratinized stratified squamous epithelium showing minimal involvement. The tumor cells exhibited intracellular cytoplasmic brown-pigment deposition, especially for the cells within the connective tissue papillae. The tumor cells proliferated deep into the oral submucosa with a

diffusely infiltrative pattern and islands of cells showed varying degrees of mitotic activity, hyperchromatism, polymorphism and anaplasia (Fig. 2A, B).

The infiltrative melanocytic cells were predominantly plasmacytoid shaped cells, arranged in both solid and alveolar patterns (Fig. 2C, D). Most of the cells were large and round to ovoid with large nuclei and prominent pink nucleoli. Brown pigmentation consistent with melanin was found both intra-and extracellularly (Fig. 2C, D).

Immunohistochemical stains for S-100, HMB-45, AE1/AE3, neuron specific enolase (NSE) and CD 45 were performed. AE1/AE3 and CD 45 were found to be negative (Fig. 3D, AE1/AE3 not shown) ruling out a carcinoma or lymphoma, whereas all others were diffusely positive (Fig. 3A, B, C). These stains helped to confirm our H & E diagnosis of oral MM.

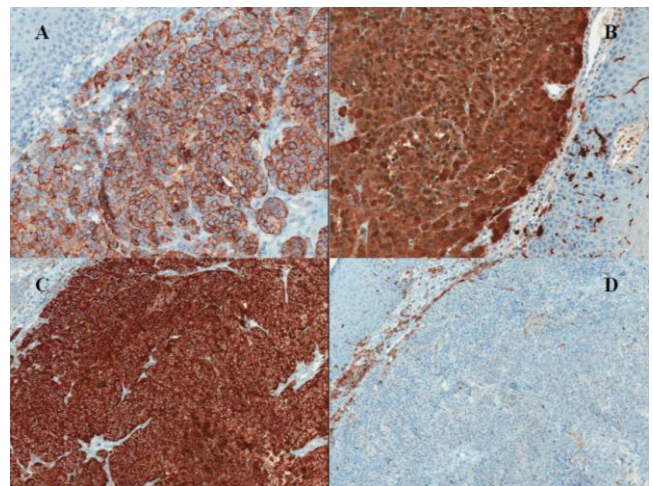


Figure 3. The neoplastic cells demonstrate diffusely positive immunoreactivity for HMB-45 (A), diffusely strong immunoreactivity for S-100 (B) and Neuron Specific Enolase (NSE) (C) indicate a significant for malignant melanoma. Absent of immunoreactivity for CD45 (D) ruling out carcinoma for this case. (Magnification x20) (A, B and C), (Magnification x10) (D).

The patient underwent a PET-CT scan showing an increasing Standardized Uptake Value (SUV) in the posterior right maxilla, (Fig.4A) as well as in nodal regions of the right neck (Fig.4B). The radiology report noted a prominent mass in the right submandibular region with a central area of decreased attenuation, inducing an extrinsic mass effect on the right submandibular gland.

Moreover, there was a second mass in the midline of the retropharyngeal region with enhancing borders and a central area of decreased attenuation compatible with a necrotic mass, most likely a lymph node, measuring approximately 2 cm (Fig.4B).

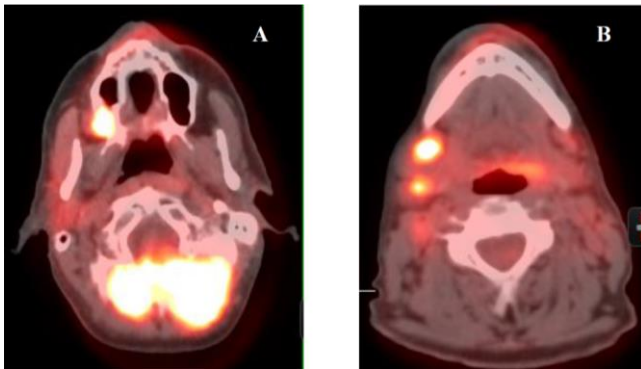


Figure 4. PET-CT showing increased SUV of the right posterior maxilla involving the right maxillary sinus (A). Additionally, there is increased SUV of right lateral neck nodes indicating metastatic disease (B).

The patient's final assessment was a clinical stage T_{4A}N₁M_x oral MM. Understanding the poor prognosis for such a lesion, localized surgical partial maxillectomy was performed for the primary oral lesion (Fig. 1B) along with ipsilateral selective neck dissection. This therapy was followed by adjuvant radiation and chemotherapy. The surgery was performed without significant incident and the patient was extubated immediately post-operatively. The patient was reconstructed with a dental obturator to optimize monitoring of the tumor's resected margins and for recurrence monitoring.

Discussion

Oral MMs can appear clinically in various forms according to their growth phases: (1) pigmented maculae; (2) pigmented plaque; (3) pigmented nodule, and these include amelanotic variants of the 3 forms.^{3,7} Unlike cutaneous melanoma, there is no effective staging classification system for oral MM. Depth of invasion has been using for determining the prognosis of skin MMs for years.⁴⁻⁸ However, within the oral cavity, depth of invasion must be given a different criteria than the traditional Breslow score. While it is still recommended to attempt to score the lesion, the thinness of the oral mucosa and rareness of oral MMs makes

correlation with prognosis much more difficult than seen in skin lesions.⁴⁻⁸

Histopathological differential diagnoses include almost any tumors. Similarly in our case, poorly differentiated carcinomas and anaplastic large-cell lymphomas or pleomorphic sarcoma must be ruled out due to the finding of large pleomorphic cells in the specimens. Immunohistochemical staining was employed in this case to highlight intermediate filaments or antigens specific for a particular cell types. HMB-45, S-100 protein, and cytokeratin (AE1/AE) are widely used for diagnosis of oral MM. However, recently use of microphthalmic transcription factor, tyrosinase and melano-A (Melan-A) immunostains have been shown to be beneficial in highlighting melanocytes as well.^{3,6}

Oral MM can also be confused with several benign lesions. The clinical differential diagnosis of many oral MM includes melanosis, nevi, melanotic macule, melanoacanthoma, racial pigmentation and/or post inflammatory pigmentation or drugs induced pigmentation. In addition, a number of syndromes or systemic diseases can induce oral pigmentation, such as Peutz-Jeghers syndrome (perioral pigmentation, intestinal polyps), LAMB syndrome (mucosal lentiginos, myxoma of the heart and skin), LEOPARD syndrome (mucosal lentiginos, Electrocardiographic conduction abnormalities, ocular hypertelorism), Addison's disease, as well as the uncommon macular pigmentation of Laugier and Hunziger disorder.^{1,8}

Surgery provides the best chance for long-term survival.^{4,9}

Decisions need to be tempered regarding use of a radical surgical approach in terms of local failure and functional outcome. However, at present, radical surgery offers the best chance of cure and local control for oral MM.⁷

The overall 5-year survival rate of gingival MM is 18% and decreasing to 11% for palatal tumors.²

Median survival rate remains at 18 months once there is lymph node involvement.²

Conclusion

In conclusion, although oral MM is extremely rare, the overall poor prognosis must encourage health care practitioners to be suspicious of any change in color or texture of oral mucosa. Early biopsy, diagnosis and

treatment of these lesions are key to the overall prognosis and morbidity of oral MM.

Declaration of Interest

The authors report no conflict of interest and the article is not funded or supported by any research grant.

References

1. Patrick, R.J., N.A. Fenske, and J.L. Messina, Primary mucosal melanoma. *Journal of the American Academy of Dermatology*, 2007. 56(5):828-34.
2. Hicks, M.J. and C.M. Flaitz, Oral mucosal melanoma: epidemiology and pathobiology. *Oral oncology*, 2000. 36(2):152-69.
3. Gu, G.M., J.B. Epstein, and T.H. Morton, Jr., Intraoral melanoma: long-term follow-up and implication for dental clinicians. A case report and literature review. *Oral surgery, oral medicine, oral pathology, oral radiology, and endodontics*, 2003. 96(4):404-13.
4. Rapini, R.P., et al., Primary malignant melanoma of the oral cavity. A review of 177 cases. *Cancer*, 1985. 55(7):1543-51.
5. Ulusal, B.G., et al., Primary malignant melanoma of the maxillary gingiva. *Dermatologic surgery : official publication for American Society for Dermatologic Surgery*, 2003. 29(3):304-7.
6. Garzino-Demo, P., et al., Oral mucosal melanoma: a series of case reports. *Journal of cranio-maxillo-facial surgery : official publication of the European Association for Cranio-Maxillo-Facial Surgery*, 2004. 32(4):251-7.
7. Femiano, F., et al., Oral malignant melanoma: a review of the literature. *Journal of oral pathology & medicine : official publication of the International Association of Oral Pathologists and the American Academy of Oral Pathology*, 2008. 37(7):383-8.
8. Barker, B.F., et al., Oral mucosal melanomas: the WESTOP Banff workshop proceedings. *Western Society of Teachers of Oral Pathology. Oral surgery, oral medicine, oral pathology, oral radiology, and endodontics*, 1997. 83(6): 672-9.
9. Ardekian, L., et al., Primary gingival malignant melanoma. Report of 3 cases. *Journal of periodontology*, 2000. 71(1):117-20.