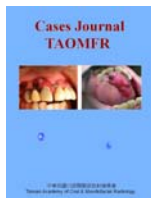


Primary Melanoma of the Maxillary Gingiva and Palate: Report of a Case

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Cases Journal TAOMFR 2011; 3:3

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Keywords: oral melanoma, palate, gingiva

Received: 17 July 2011; Accepted: 28 July, 2011

Melanoma is a malignant neoplasm arising from melanocyte, which originates from neural crest. Weber presented the first reported case of primary oral melanoma in 1859 [1, 2]. Oral melanoma is an infrequent neoplasm making up less than 1% of all melanomas. Intraoral melanomas are highly malignant and the median survival was 2 years and the 5-year survival rate was 25% [3]. The peak age is between 40 and 60 years. Male is slightly more frequent affected; nearly 50% of these melanomas are on the hard palate and about 25% are on the maxillary gingiva. Intraoral melanomas are preceded by an area of hyperpigmentation which vary from black to brown, often by many years. Intraoral melanoma may be flat, but arise usually raised or nodular and asymptomatic initially, ulceration, painful or bleeding may be seen later. Histopathologically, malignant melanocytes invade both epithelium and connective tissue. Growth of melanoma is radial rather than invasive, and there may be a few scattered melanocytes in the superficial corium associated with a spares inflammatory cellular infiltrate.

CASE PRESENTATION

A 25-year-old female complained of a swelling mass over her anterior palatal gingiva for about 3 years. She went to local dental clinic for excision of the mass for several times since August 2005 to May 2008. Since the mass kept multiple recurring and enlarging, she visited our dental clinic for further help. Oral examination revealed black pigmentation deposition over maxillary

gingiva extending from tooth 13 to 23 area (Figure 1A) and an erythematous with black pigmentation rubbery, nodular appearance mass over palatal gingiva over tooth 15 to 22 area (Figure 1B). Temporary resin bridges were fabricated on tooth 13 to 23. Multiple missing teeth were noted. The left mandibular second and third molars revealed severe mesial titling.

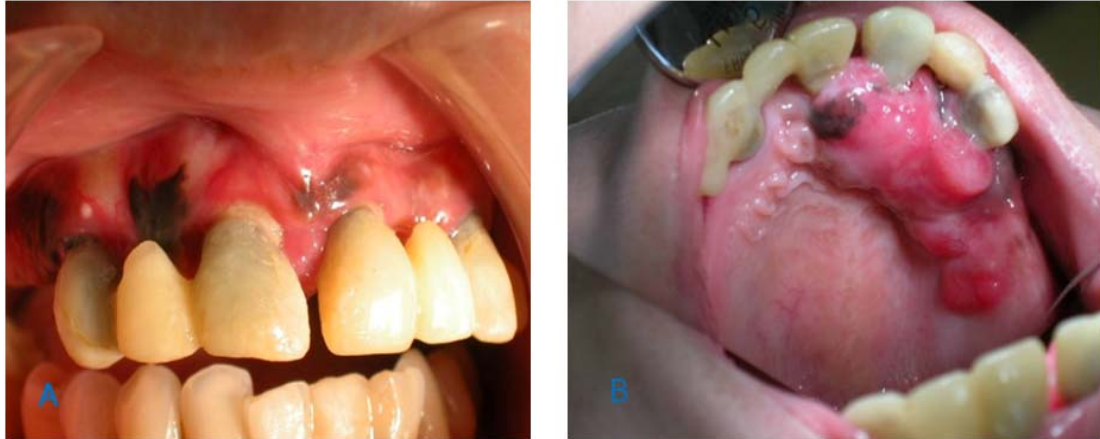


Figure 1 Buccal aspect (A) of the lesion revealed hyperpigmentation over right maxillary gingiva; palatal aspect (B) revealed an erythematous with black pigmentation rubbery, nodular appearance mass over palatal mucosa from tooth 15 to 22 area

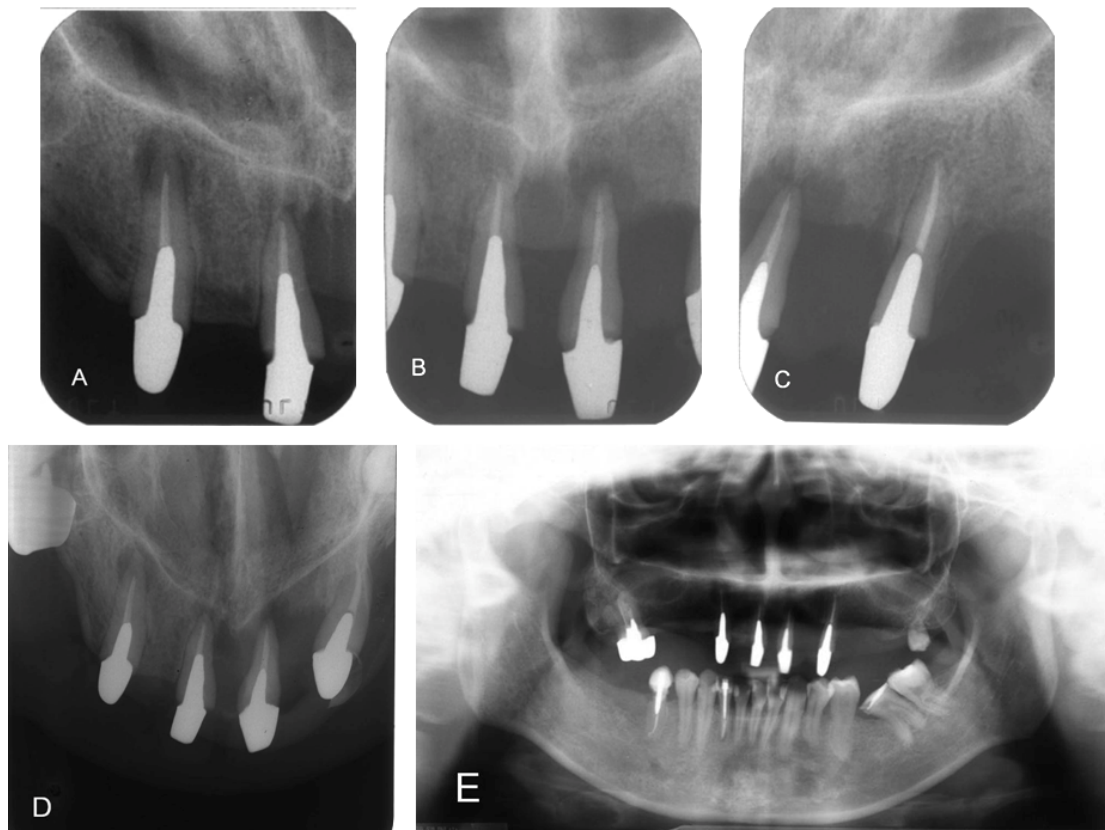


Figure 2 Intraoral periapical (A-C) and occlusal radiographs (D) of the maxillary anterior teeth revealed alveolar bony destruction over tooth 11 and 21 and surface erosion of the region of edentulous alveolus over tooth 22 and 24 area. Incomplete endodontic fillings with periapical lesions over tooth 11, 13, 21 and 42 were seen in periapical (A-C) and panoramic (E) radiographs

Incomplete endodontic fillings with periapical lesions over tooth 11, 13 and 21 in periapical radiographs (Figure 2A-C). Irregular alveolus destruction of tooth 11 and 21 area and surface erosion

of the edentulous alveolar bone of tooth 12 and 14 were seen in intraoral periapical (Figure 2A-C), occlusal (Figure 2D) and panoramic radiographs (Figure 2E).

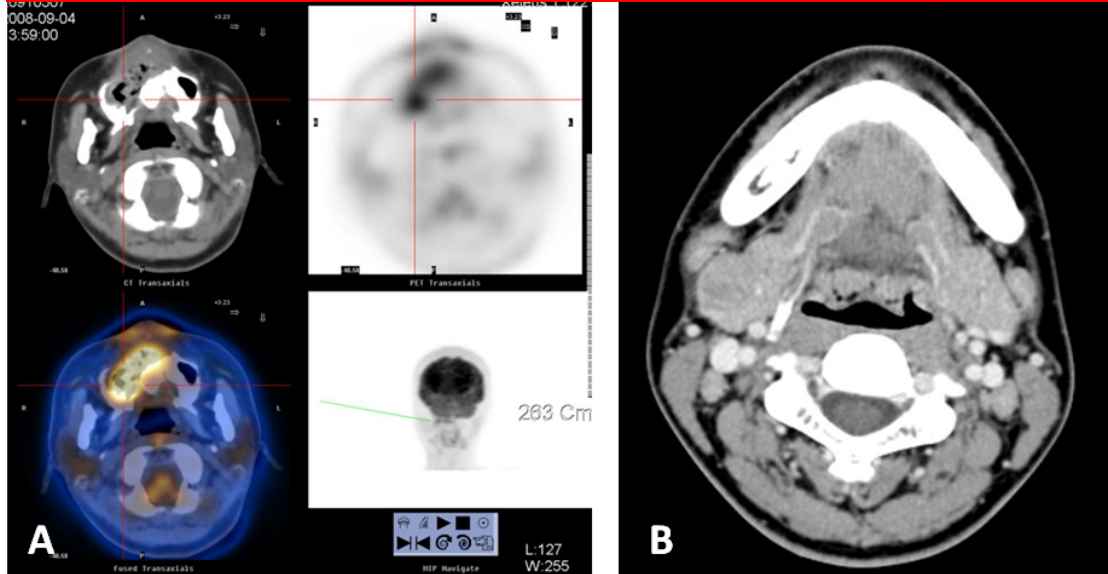


Figure 3 Computed tomography-positron emission tomography image revealed a soft tissue nodule in right upper gingiva with maxillary alveolar process invasion (A) and a nodule at right cheek subcutaneous region anterior to right mandible (B)

Computed tomography-positron emission tomography (CT-PET) revealed a soft tissue nodule about 1.8 cm in diameter in right upper gingiva with maxillary alveolar process invasion, consistent with a tumor growth (Figure 3A). There was a round nodule about 1.8 cm in diameter with relatively low enhancement in right submandibular gland (Figure 3B). In addition, there was a suspicious nodule about 1.1 × 0.6 cm

with contrast enhancement at right cheek subcutaneous region anterior to right mandible. Presence of borderline size lymph nodes at bilateral neck level IB and nodal metastases was suspected. Scintigraphic findings suggested bony metastasis (Figure 4) and CT-PET revealed a low enhancement mass in right lung (Figure 5). Distant metastasis of the lesion was hence strongly suspected.

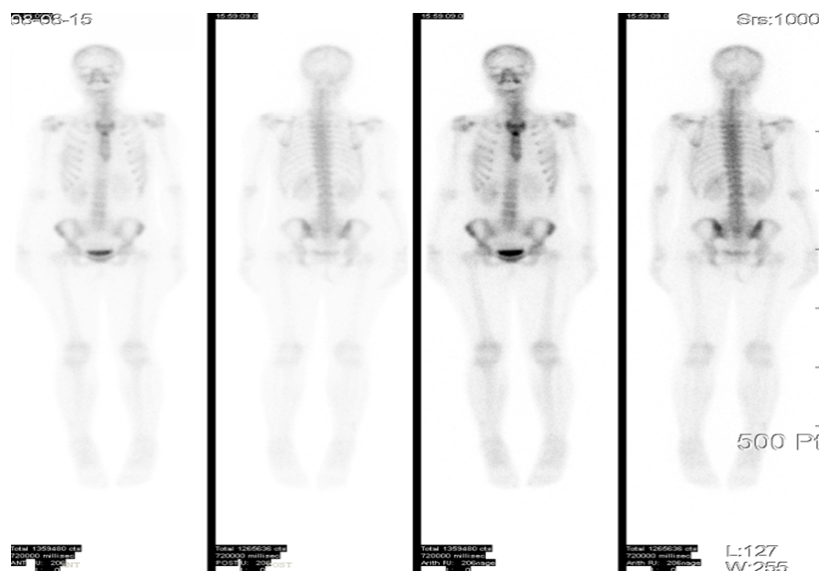


Figure 4 Scintigraphic findings suggested periodontal disease and/or bony metastasis

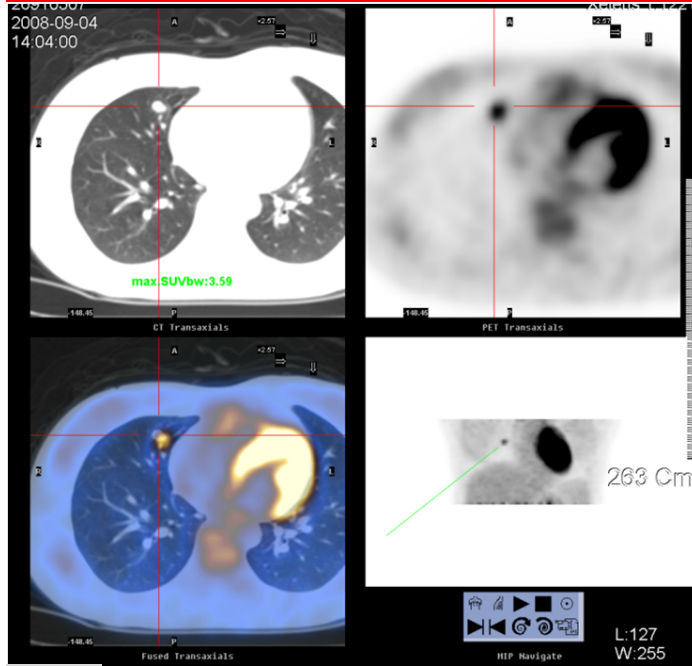


Figure 5 Distant metastasis of right lung was suspected for chest and abdomen scan

An incisional biopsy of the lesion was done under local anesthesia and the specimen taken from the palatal gingiva and was sent for histopathological examination. Hematoxylin-eosin stained section revealed round, spindle-shaped cells infiltrated in the connective tissue stroma. Sheets of the epithelioid cells were intermingled with spindle-shaped

cells showing sarcomatous, swarming pattern (Figure 6A and B). Pleomorphic, hyperchromatic cells revealed cytoplasmic stainings with S-100 (Figure 6C) and HMB-45 (Figure 6D). Finally, histopathologically, the lesion showed melanoma of palatal gingiva. Unfortunately, the patient was subsequently lost follow-up.

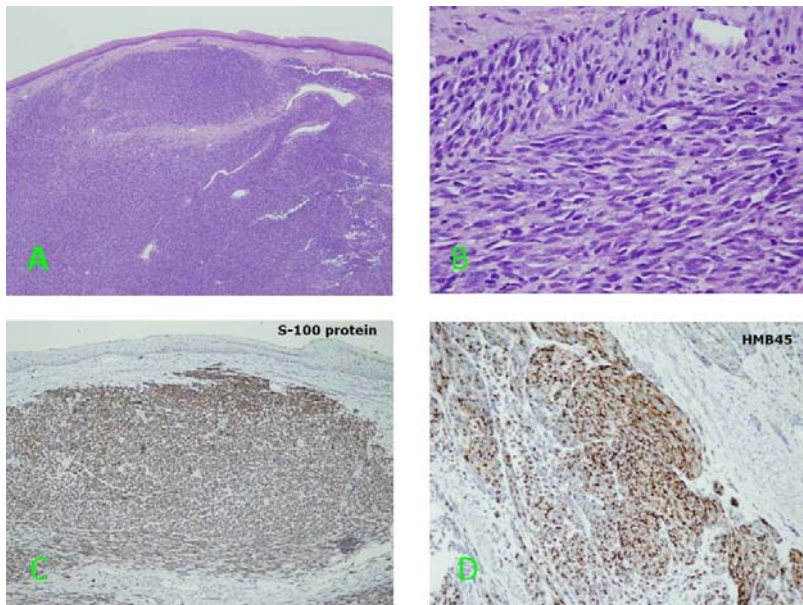


Figure 6 Hematoxylin-eosin stain (A and B) revealed nests of round and spindle-shaped dysplastic cells infiltrated in the connective tissue stroma; immunohistochemical staining of S-100 (C) and HMB-45 (D) showed diffuse positivity in the tumor cells (A ×40, B ×400, C ×40, D ×100)

COMMENTS

Oral melanomas exhibit much more aggressive behavior than those found on the skin. The predisposing factors for the cutaneous melanomas are either ultraviolet radiation or have cytogenetic defects [4]. Although some primary oral melanomas are supposed to occur either from nevus, pre-existing pigmented areas or *de novo*, no well-established etiologic or risk factors have been identified for intraoral melanomas. Mechanical traumas including injury from ill-fitting prostheses as well as infection to the oral mucosa have been suggested as possible factors, but all still lack direct proof for the etiological role. Although our patient had excised the lesion for many times since August 2005, no histopathological examination proved whether this melanoma was transformed from previous lesion or not.

Anaplastic, non-pigmented malignant melanoma can be confused with other mesenchymal tumors, and can be sarcoma-like. Hence, correct diagnosis has been greatly helped by immunohistochemistry, such as S-100, melanoma-associated antigen (MMA) and HMB-45 positivity [5]. Therefore, Barrett [6] suggested that intraoral malignant melanoma, on the basis of its clinicopathologic as well as immunohistochemical features, should be regarded as a separate pathological entity from cutaneous melanoma.

Panoramic radiograph is the most often used radiological imaging technique for jaw bone lesions, but this technique is unable to identify the soft tissue tumors as well as cervical lymph node involvement. PET with fluorodeoxyglucose (FDG) is an established imaging tool for the assessment of patients with head and neck cancer and is able to identify the primary tumor as well as regional lymph node involvement [7-11]. Intraoral

melanomas can be visualized using FDG-PET, as demonstrated in our case. Furthermore, locoregional and distant metastases can be evaluated much like those of cutaneous malignant melanoma [12]. Therefore, PET may be suitable for the staging and/or restaging of these patients. Moreover, PET should be acquired in a whole body mode, i.e. including the chest and abdomen in all patients with malignant tumor of the oral cavity [13-15]. In addition to the primary intraoral lesion, a suspicious nodule over right cheek subcutaneous region anterior to right mandible and enlarged lymph nodes at bilateral neck level IB could be seen in head and neck CT/PET. Furthermore, distant metastasis of the right lung was suspected in the chest and abdomen scan for our patient.

CONCLUSION

The current case has been unfortunate for delayed histological diagnosis. Although the previous intraoral lesion was excised once again, no specimen was sent for histopathological examination to confirm the diagnosis. Therefore, we emphasized that general practice dentists should be alerted for the intraoral lesions with rapid growth and those lesions should be subjected to histopathological examination. Finally, we also recommend that PET image is useful for diagnosis of intraoral primary melanoma with locoregional lymph node involvement and distant metastases. ■

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This article has been peer reviewed

REFERENCES

1. Greene, GW, Haynes JW, Dozier M, Blumberg JM, Bernier JL. Primary malignant melanoma of the oral mucosa. *Oral Surg Oral Med Oral Pathol* 1953;**6**:1435-43.
2. Robertson GR, DeFiebre BK, Firtell DN. Primary malignant melanoma of the mouth. *J Oral Surg* 1979;**37**:349-52.
3. Chiu NT, Weinstock MA. Melanoma of oronasal mucosa. Population-based analysis of occurrence and mortality. *Arch Otolaryngol Head Neck Surg* 1996;**122**: 985-8.
4. Silverman S. Oral Cancer. 2003, BC Decker Inc: Hamilton London. p. 155-7.
5. Gazit D, Daniels TE. Oral melanocytic lesions: differences in expression of HMB-45 and S-100 antigens in round and spindle cells of malignant and benign lesions. *J Oral Pathol Med* 1994; **23**:60-4.
6. Barrett AW, Bennett JH, Speight PM. A clinicopathological & immunohistochemical analysis of primary oral mucosal melanoma. *Eur J Cancer B Oral Oncol* 1995; **31B**:100-5.
7. Rege S, Maass A, Chaiken L, Hoh CK, Choi Y, Lufkin R, et al. Use of positron emission tomography with fluorodeoxyglucose in patients with extracranial head and neck cancers. *Cancer* 1994;**73**:3047-58.
8. Laubenbacher C, Saumweber D, Wagner-Manslau C, Kau RJ, Herz M, Avril N, et al. Comparison of fluorine-18-fluorodeoxyglucose PET, MRI and endoscopy for staging head and neck squamous-cell carcinomas. *J Nucl Med* 1995;**36**:1747-57.
9. Wong WL, Chevretton EB, McGurk M, Hussain K, Davis J, Beaney R, et al. A prospective study of PET-FDG imaging for the assessment of head and neck squamous cell carcinoma. *Clin Otolaryngol Allied Sci* 1997;**22**:209-14.
10. McGuirt WF, Williams DW 3rd, Keyes JW Jr, Greven KM, Watson NE Jr, Geisinger KR, et al. A comparative diagnostic study of head and neck nodal metastases using positron emission tomography. *Laryngoscope* 1995; **105**:373-5.
11. Benchaou M, Lehmann W, Slosman DO, Becker M, Lemoine R, Rufenacht D, et al., The role of FDG-PET in the preoperative assessment of N-staging in head and neck cancer. *Acta Otolaryngol* 1996;**116**:332-5.
12. Goerres GW, Stoeckli SJ, von Schulthess GK, Steinert HC. FDG PET for mucosal malignant melanoma of the head and neck. *Laryngoscope* 2002;**112**:381-5.
13. Wax MK, Myers LL, Gabalski EC, Husain S, Gona JM, Nabi H. Positron emission tomography in the evaluation of synchronous lung lesions in patients with untreated head and neck cancer. *Arch Otolaryngol Head Neck Surg* 2002;**128**:703-7.
14. Kitagawa Y, Nishizawa S, Sano K, Sadato N, Maruta Y, Ogasawara T, et al. Whole-body (18)F-fluorodeoxyglucose positron emission tomography in patients with head and neck cancer. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2002;**93**:202-7.
15. Jackel MC, Rausch H. Distant metastasis of squamous epithelial carcinomas of the upper aerodigestive tract. The effect of clinical tumor parameters and course of illness. *HNO* 1999;**47**:38-44.