Oral Malignant Melanoma: A Rare Case Report and Review

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ABSTRACT
Primary malignant melanoma is only rarely found in oral cavity (estimated between 0.2 and 8.0% of all melanomas) and occurs approximately four times more frequently in oral mucosa of the upper jaw, usually on the palate, alveolar gingiva and melanoma of mandibular gingiva is extremely rare. The peak age of diagnosis of melanoma is between 55 and 65 years. A biopsy is required to confirm the diagnosis. Here, we present a rare case of malignant melanoma of mandibular gingiva in a 64-year-old female patient which is confirmed by histopathology.

Keywords: Bone destruction, 3D reconstruction CT scan, Female incidence, Mandibular gingiva.

INTRODUCTION
Malignant melanoma is a neoplasm of melanocyte origin that arises from a benign melanocytic lesion or de novo from melanocytes within otherwise normal melanoma or skin. It was reported by Weber in 1859 and is considered to be the most biologically unpredictable and deadly of all human neoplasms. Over 90% of the melanomas occur on skin surfaces with slightly over 1% of melanomas arising from the mucosal surfaces. The most common sites for mucosal melanomas are the head and neck region (55%), followed by anal and rectal region (24%), female genital tract (18%) and urinary tract (3%). Head and neck melanomas of mucosal origin account for less than 1% of all melanomas with an incidence of oral mucosal melanomas being 1.2 cases per 10 million population per year, which accounts for about 0.2 to 8.0% of all reported melanomas. The etiology of oral mucosal melanomas is unknown, tobacco abuse and chronic irritation from ill-fitting dentures being mentioned as possible risk factors. It is largely a disease of those older than 40 years with a male to female ratio of 2:1 and it often occurs on hard palate, maxillary gingiva. Its occurrence on mandibular gingiva is extremely rare.

This clinical entity which is the rarest of its kind being mucosal and present on mandibular gingiva is being reported here.

CASE REPORT
A 64-year-old female patient reported to the Department of Oral Medicine and Radiology, PMNM Dental College and Hospital, Bagalkot, Karnataka, India, with a chief complaint of growth in the left lower posterior region of mandible since 1 month which gradually increased in size and associated with dull, intermittent, localized type of pain which relieved temporarily on taking analgesics. The patient’s medical history revealed uterine cervix cancer 5 years back for which she had undergone radiotherapy. Dental and family history was noncontributory.

Extraoral examination did not reveal any abnormality. Single submandibular lymph node was palpable which was of 1.5 to 2 cm in size, nontender, firm in consistency but not fixed.

On intraoral examination, there was an exophytic proliferative mass of size 2.5 × 4 cm extending buccolingually from buccal vestibule to lingual vestibule and mediolaterally from medial aspect of mandibular left first premolar to medial aspect of mandibular left second molar (Fig. 1). The mucosa over the growth was rough, lobulated with well-defined margins

Fig. 1: Intraoral photograph showing a soft tissue pedunculated swelling
and the color ranged from dark bluish to black. The surrounding hard and soft tissues were normal. On palpation, the mass was pedunculated, tender, firm in consistency with bleeding tendency and the teeth within the growth (i.e. first and second premolars) showed grade II mobility. There were no other pigmented areas in oral mucosa.

The intraoral periapical radiograph and orthopantomograph showed periodontal ligament thickening with respect to mandibular left first and second premolar, second molar, with irregular bone destruction and erosion extending from first premolar to second molar region with floating teeth appearance (Fig. 2). Chest radiograph did not show any metastatic foci. Computerized tomography scan (CT scan) examination revealed 21 × 11 mm lobulated enhancing hyperdense mass in left lower gingivobuccal sulcus suggesting a malignant tumor (Fig. 3A). 3D reconstruction CT scan showed erosion on bone in left mandibular premolar and molar region (Fig. 3B).

Blood pictures (complete blood count, erythrocyte sedimentation rate) were within normal limits and peripheral smear showed microcytic hypochromic anemia and relative neutrophilia. Liver function tests were normal with slight elevation of alkaline phosphatase levels.

The lesion was subjected to incisional biopsy and the histopathology on hematoxylin and eosin stained section revealed loose fibrous connective tissue predominantly consisting of nodular masses of lesional cells which were large, round with granular cytoplasm (melanin granules) arranged in sheets and islands (Fig. 4). These features were consistent with malignant melanoma.

**DISCUSSION**

Oral malignant melanoma is a rare aggressive neoplasm comprising of melanocytes. The incidence of primary oral malignant melanomas has been reported to be between 0.2 and 8% of all melanomas. It has been estimated that 1 in 75 persons born in the year 2000 will develop cutaneous melanoma in his lifetime. Unlike cutaneous melanomas, oral lesions are more common in men than women, which is in contrast to present case which occurred in a woman and hence is a rare incidence. The most common site being hard palate and maxillary gingiva, it is extremely rare in mandibular gingiva and this rare occurrence on mandibular gingiva is being reported in the present case report. Currently, most melanomas are thought to arise de novo, although the roles of inhaled and ingested carcinogens as from tobacco use and chronic irritation from ill fitting dentures in pathogenesis have been suggested.

Ingested and inhaled environmental carcinogens at high body temperature may play some role. In present case, patient did not have history of any habit or any other chronic irritation except local irritants like calculus and stains. Our patient had a history of treatment for cancer of the cervix. It is therefore thought that some primary tumors can regress completely, but leave their metastases behind. Vasculogenic mimicry has been seen in melanoma. Anoikis has been suggested to act as a physiological barrier to metastasis; resistance to anoikis may allow survival of cancer cells during systemic circulation, thereby facilitating secondary tumor formation in distant organs.

Melanoma may present itself in two patterns, in the first pattern there is rapid appearance and enlargement of pigmented lesion, as in our case and in second, the tumor is preceded by pigmented area for a variable period. Most lesions appear as asymmetric pigmented maculae of irregular shape, they may grow in horizontal direction for some years prior to a phase of...
vertical growth and submucosal invasion. The progress of melanoma may go through three phases; a nodular phase usually affecting the center, a flat or slightly elevated deep brownish-black pigmented plaque phase and a nonelevated light-brown macular phase, in the present case the lesion was in nodular phase.

At presentation approximately 13 to 19% of patients have lymph node metastasis and another 16 to 20% are likely to develop metastases subsequently. Hence, regional lymph node involvement represents controversial issue. According to Umeda and Shimada (1994), chance of cervical node involvement increases when the thickness of an oral melanoma reaches 5 mm or more. Conley and Pack in 1974 found lymph node metastases from mucous membrane melanomas to be less frequent than cutaneous lesions. Our present case did not reveal lymph node involvement. The so-called ABCD checklist (asymmetry, border irregularities, color variegation and diameter >6 mm) that is commonly used in identification process of cutaneous melanoma could also be of some help in diagnosis of oral melanoma. Tanaka et al classified oral melanomas into five types based on their clinical appearance as pigmented nodular, nonpigmented nodular, pigmented macular, pigmented mixed and nonpigmented mixed type. Among these, the present case was of pigmented nodular type and also fulfilled ABCD checklist.

Clinical Staging System for Oral Malignant Melanoma with Histopathological Microstaging

- **Stage I:** Primary tumor present only (Tany N0 M0)
  - Level I—pure in situ melanoma without evidence of invasion or in situ melanoma with microinvasion
  - Level II—invasion up to lamina propria
  - Level III—deep skeletal tissue invasion into skeletal muscle, bone or cartilage
- **Stage II:** Tumor metastatic to regional lymph nodes (Tany N1 M0)
- **Stage III:** Tumor metastatic to distant sites (Tany Nany M1)

Our case had invasion into connective tissue histologically and clinically invasion into the underlying bone. Hence, the melanoma is in stage I level III.

Involvement of jaw bones by primary and secondary melanoma radiographically is very rare. However, when they do involve the bone they are indistinguishable from osteomyelitis, while others have appearance found with any other lytic malignant tumor. Radiographically, melanomas show ill-defined radiolucency with invasive border and floating teeth appearance. Our case presented with similar features on radiographs. Chest radiograph showed no metastatic foci. CT scans depict malignant melanoma as an expansile, homogeneously enhancing mass. CT scan of the present case revealed lobulated enhancing hyperdense expansile mass suggestive of a malignant tumor.

The differential diagnoses of pigmented lesions of oral mucosa include tattoo (amalgam, graphite), oral melanotic macule, nevi, melanoacanthoma and melanoma. When an oral pigmentation cannot be confidently diagnosed as benign on clinical grounds, a biopsy is mandatory in order to exclude oral malignant melanoma. The size of the lesion or anatomic limitations may preclude the taking of an excisional biopsy in the oral cavity, but an incisional biopsy can be taken through the thickest or the most suspicious part of the tumor, in case of large lesion. A nomenclature was proposed at the 1995 WESTOP Banff workshop to classify oral melanomas by histological pattern as in situ, invasive and combined in situ and invasive. The present case on incisional biopsy showed both in situ and invasive histological pattern. Microscopically, melanomas in situ show an increase in atypical melanocytes. These atypical melanocytes have angular and hyperchromatic nuclei, they may form aggregates or may be irregularly distributed in a junctional location. The melanocytes present in invasive melanomas show a variety of cell types, including epitheloid, spindle and plasmacytoid, they typically have large vesicular nuclei with prominent nucleoli, mitoses may be present, but usually not in large numbers, they are usually aggregated into sheets or alveolar groups.

Radical surgery is the treatment of choice for oral melanoma, elective neck dissection has also been advocated. Adjuvant chemotherapy with decarbazine, platinum analogs, nitrosureas and microtubular toxins have been used for palliative purposes or for therapy of metastatic melanoma, but does not seem to influence survival. Systemic immunotherapy has been used as adjuvant or for palliation in the treatment of disseminated disease. Radical surgery in combination with radiotherapy and chemotherapy or radiotherapy alone is preferred in inoperable tumors or in elderly. Our patient underwent radical surgery followed by radiotherapy. Local multiple recurrences are common cause of failure. However, our case presented with negative nodes, hence, recurrence is less likely as stated by MJ Hicks and CM Flaitz. Prognosis although poor is highly variable with oral melanomas; 5-year survival is 15% with a median survival of 25 months. Gingival melanoma (18%) has slightly greater...
5-year survival than for palatal melanoma (11%) with a considerably longer median survival period (46 months vs 22 months).\(^3\) Prevention and screening for mucosal melanomas of head and neck involve annual evaluation for pigmented lesions in the oronasal and upper respiratory tract.\(^3\) The best likelihood for favorable outcome is early detection and excision of melanocytic mucosal lesions.\(^3\)

**CONCLUSION**

Despite improvement in surgical techniques and the introduction of new chemotherapeutic agents, prognosis of this malignancy remains poor. Oral malignant melanoma may demonstrate significant heterogeneity in morphological features, developmental process and biological behavior,\(^{12}\) hence early diagnosis is essential for successful treatment and more cases are required to be reported for better understanding of variations in presentation of this tumor.

**REFERENCES**