Malignant melanoma of lingual gingiva with ipsilateral submandibular lymph node metastasis: a case report

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Abstract Malignant melanoma is a neoplasm of epidermal melanocytes. It is one of the most biologically unpredictable and deadly of all human neoplasms. However, malignant melanoma in the oral cavity is a rare malignancy, accounting for 0.2% to 8% of all melanomas. It has a grave prognosis, with a 5 year survival of 10–20%. We present a case of malignant melanoma of lingual gingiva of left mandibular molars with ipsilateral submandibular lymph node metastasis. We performed peripheral osteotomy of primary lesion followed by modified radical neck dissection by sparing internal jugular vein and patient had received postoperative radiotherapy.

Keywords Malignant melanoma · Lymph node metastasis

Introduction

Oral cancer accounts for approximately 3.5% of all human cancers, of which approximately 96% are carcinomas. Malignant melanoma is the third most common cancer of skin (Basal and Squamous cell carcinomas are more prevalent). It accounts for only 3% of all such malignancies. Mucosal melanoma was first described by WEBER in 1859 [1,2].

Approximately 17,400 new cases of melanoma were diagnosed in the United States. About 20% of all melanomas occur in head and neck area. The frequency with which melanoma is diagnosed is rapidly increasing in almost all parts of the world. The reasons for the increased number of cases are not clear but may be related to increased recreational activity in the sun, changes in clothing migration to the 'Sunbelts', and an increase in ultraviolet light exposure from a decrease in atmospheric ozone layer [3].

Case report

A 55-year-old male patient reported to the Department of Oral and Maxillofacial surgery, Sibar Institute of Dental Sciences with 8-month's history of a swelling in left

sub-mandibular region (Fig. 1). The past medical history and past surgical history were non contributory. Social history was positive for 25 years of smoking cigarettes and alcohol intake. Extraoral examination revealed a well defined, oval, non tender, fixed, and hard lymph node swelling of size 4x3 cm on left sub-mandibular region. Intraorally black macular discoloration of lingual gingiva of size 3x1 cm extending from mandibular left first molar to left 3rd molar region (Fig. 2).

Hematological and biochemical investigations were normal. CT scan showed no mandibular bony changes. On aspiration of enlarged lymph node, dark brown to black colored material was obtained. FNAC revealed few pleomorphic cells with round to polygonal shape. Cytoplasm was eosinophilic, containing fine dark brown melanin pigment material. In most of the cells, this brown pigment was obscuring the nuclei. Suggestive of metastasis from melanoma.

An incision biopsy of mandibular lingual gingival lesion revealed an atypical melanocytic proliferation with moderate atypia, features consistent with malignant melanoma (Fig. 3). We performed peripheral osteotomy of intraoral lesion followed by modified radical neck dissection on the left side sparing the

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internal jugular vein (Figs. 4, 5 and 6). The patient had received a total dose of 3000 cGY of radiotherapy in 15 fractions within a 3 week period.

Discussion

Melanomas are malignant neoplasms arising from melanocytes originating from the neural crest. The cells and the corresponding neoplasms arising from the neural crest are grouped under the Dispersed Neuro Endocrine System (DNES), which were previously designated as Amine Precursor Uptake and Decarboxylase (APUD'oma) tumors [1]. Melanocytes are primarily present in the basal portion of epidermis at the dermo-epidermal junction. Primary malignant melanoma has been described in virtually all sites and organ systems where neural crest cells migrate.

The incidence of primary oral malignant melanomas has been reported to be between 0.2 and 8% of all melanomas (Pliskin, 1979). Rapini (1985) reviewed three large studies of oral malignant melanoma (Chaudhry et al. 1958; Takagi et al. 1974; Liversedge, 1975) along with his own study of 177 cases and found that over 82% of the patients were over 40 years and 37% were more than 60

Fig. 1 Left submandibular lymph node swelling

Fig. 2 Black melanotic plaque on lingual gingiva

Fig. 3 Incision biopsy of lingual gingival lesion showing atypical melanocytic proliferation (Hematoxylin and Eosin; Original magnification X10)

Fig. 4 Modified radical neck dissection with preservation of L.J.V. and peripheral osteotomy of mandibular segment

Fig. 5 Osteotomy segment containing melanotic plaque

Fig. 6 Gross section of submandibular lymph node showing melanin pigment

years of age [4]. Unlike cutaneous melanomas, the oral lesions are more common in men than in women. The most common site for oral melanoma is the palate (32%), followed by maxillary gingiva (16%), other common sites are the mandibular gingiva (7%) lips (7%) and alveolar gingiva (5%) [2,4]. The etiologic factors for oral melanoma have not been studied extensively, although factors such as tobacco use and formaldehyde exposure have been implicated [1,4].

The presentation of oral melanoma varies. One third of patients are asymptomatic at diagnosis. The lesions may be incidentally observed by the patient or dentist. This tumor may present itself in two different patterns. In the first, there is rapid appearance and enlargement of a pigmented lesion, whereas in the second, the tumor is preceded by pigmented area for a variable period [5]. There are 3 growth phases: 1. A macular phase consisting of proliferation of dendritic melanocytes without apparent atypia and simple hyper pigmentation in the basal cell layer and melanin incontinence; 2. A pigmented plaque phase consisting of preinvasive tumor cell nests in the lower epithelial layers; and 3. A nodular phase consisting of spindle - shaped or epithelioid tumor cells in the submucosa [6]. In the present case the lesion was found to be in the nodular phase (Fig. 4).

The lesions are histologically indistinguishable from their cutaneous counterparts (Chaudhry et al. 1958) but unfortunately the histological staging of Clark's levels (Clark et al. 1975) which is useful prognostic indicator in cutaneous melanoma does not have any relevance to tumors of oral cavity due to different histological structure of mucosa and skin [3]. A nomenclature was proposed by at the 1995 WESTOP Banff workshop to classify oral melanoma by histologic pattern as in-situ, invasive, and combined in-situ and invasive [7,8]. The present lesion showed both insitu and invasive histological pattern. Special staining techniques like Fontana silver stain and the Prussian blue stain are useful in about 75% of cases and the immunocytochemical diagnosis of malignant melanoma has become the method of choice [9,10]. The demonstration of neuronal specific S-100 protein is a useful diagnostic indicator, especially if the tumor is of the amelanotic type. More recently the application of monoclonal antibody techniques (HMB-45) has increased the specificity of immunohistochemical diagnosis [7,11,12]. The ultra structural examination of melanoma by electron microscopy is an important diagnostic tool and provides specific appearances such as the presence of pre-melanosomes and melanosomes [13].

It is generally agreed that surgery is the treatment of choice for oral malignant melanoma with wide local excision and lymph node dissection of involved lymph nodes [14]. For many years oral mucosal malignant melanoma has been considered to be resistant to radiotherapy but cases of temporary regression (Curran and Whittaker, 1973) and also apparent cure (Pearson, 1974) are known to occur [2]. Chemotherapy may be used to reduce the size of the tumor preoperatively and to improve surgical management. The first line of chemotherapeutic agents is Dimethyltriazeno Imidazole Carboxamide (DTIC) alone or in combination with vincristine or dactinomycin [1]. Immunotherapy is based on the possession of specific melanoma related antigens. Immunotherapeutic drugs include interferon (INF), interleukin 2, and intralesional lymphokines. The use of INF- γ and cimetidine in combination is believed to activate killer T-cells and inhibit suppressor T-cells, resulting in a reduction of the size of melanoma [2].

Conclusion

Due to the aggressive and unpredictable nature of malignant melanoma, early identification and aggressive surgical

approach help in improving the prognosis. The 5-year survival rate for patients with melanoma of oral cavity remains poor and has been reported to fall within a range of 5–20% [2,8,11,13,14].

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