CASE NOTE

Primary malignant melanoma of the tongue

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Primary malignant melanoma of the oral cavity is a rare neoplasm. The incidence of oral cavity melanoma is about 0.2%–8.0% of all malignant melanoma cases. Primary lesions arising from oral mucosa occur most frequently on the maxillary gingiva and palate, with the lips being the third most frequent. Melanoma of the tongue is specifically uncommon and represents less than 2% of all oro-nasal melanoma cases.[1] A review of literature revealed fewer than 30 reported cases of primary malignant melanoma of the tongue.

Case report

A 73-year-old woman presented with a painless mass on the right side of her tongue that had lasted for 6 months. It started as a small nodule that progressively increased in size, especially over the previous 2 months when ulcerative changes started to occur. On clinical examination, we found a black pigmented ulcerated mass measuring about 3 × 2 cm on the right postrolateral aspect of the tongue (Fig. 1). There were no other similar cutaneous lesions intraorally or elsewhere on her body.

Computed tomography (CT) scans of her head and neck showed a right postrolateral tongue lesion with no substantial cervical lymphadenopathy. Metastatic work up showed no signs of distant metastatic disease.

We performed wide local excision of the tumour and right functional neck dissection. The histopathological findings showed malignant melanoma of the tongue, characterized by neoplastic proliferation of epithelioid to spindle melanocytes, with melanin deposits and underlying skeletal muscle invasion. Scattered tumour cell nests were also present in the overlying squamous epithelium, suggesting that the tumour was a primary rather than a metastatic lesion (Fig. 2 and Fig. 3).

The surgical resection margin and base of the tumour were negative for tumour cells, and there was no evidence of metastatic nodal disease in the neck dissection specimen. The patient had an uneventful recovery and is currently underdergoing regular follow-up.

Discussion

The mucosal membranes are rare sites for primary malignant melanoma. The presence of melanocytes in the mucosal membrane of respiratory, alimentary and urogenital tracts explains the occurrence of malignant melanoma in these sites.[2] Melanoma of the oral cavity mucosa is a distinctly rare occurrence with an incidence of 0.012 in 105 for combined primary and metastatic lesions of the oral cavity.[1]

The tumours are commonly found in patients older than 40 years, and there are no clinically important differences between the sexes.[1] The oral cavity may be a site of predilection for melanomas in Japanese people,[3] although it is very rare in white people. Oral pigmentation precedes the development of

malignant melanoma in about one-third of patients. Takagi and colleagues[3] reported that mucosal melanosis was associated with oral melanoma in 66%, pre-existing in 36.2% and concurrent in 29.8% of patients.

Oral melanomas may present as flat, painless, dark brown or black discolored macules or nodules, sometimes with erythema or ulceration. As the disease progresses, bony erosion is common. Whether the lesion is a primary malignancy or a secondary one from an occult cutaneous tumour, the distinction between them will affect the management decision and outcome.

By histopathology, Billings and colleagues[4] found that all metastatic lesions lacked evidence of junctional activity in the overlying mucosa and showed no epidermal migration. This is in contrast to primary lesions, in which 44% and 38% had junctional activity and epidermal migration, respectively. A unique feature seen in the primary lesions (25%) was the presence of extensions of the melanotic pigment into the minor salivary glands.[4]

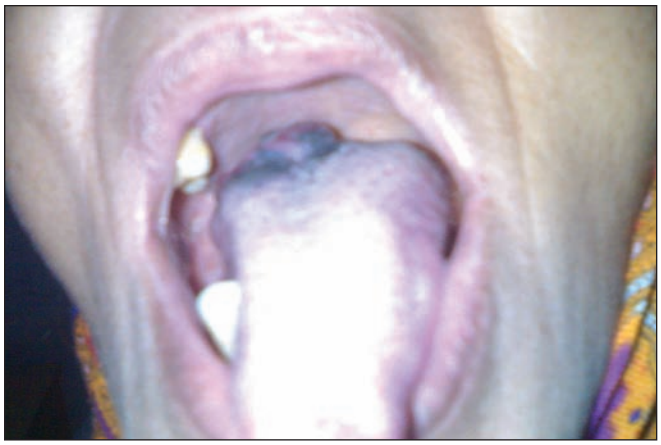

Fig. 1. We found a black, pigmented and ulcerated mass measuring about 3 cm on the right postrolateral aspect of the tongue of a 73-year-old woman.

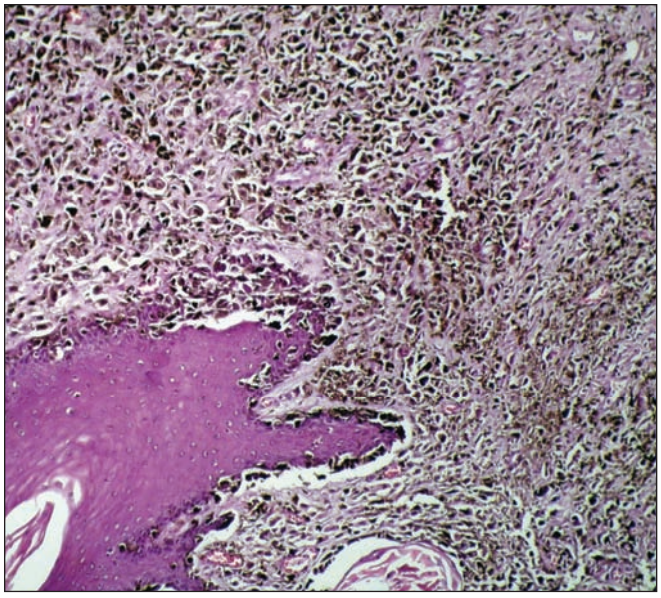

Fig. 2. Malignant melanoma of the tongue. This field exhibits radial growth of anaplastic melanocytes with evident melanin pigments in the cytoplasm in sheets of dispersed single cells directly beneath surface squamous epithelium, pagetoid spread of tumour cells into squamous epithelium and at the junction between epithelium and subepithelium (hematoxylin and eosin staining, original magnification ×100).

The immunohistochemical profile of oral malignant melanoma was similar to that of cutaneous melanoma, with the exception that no oral malignant melanoma was positive for cytokeratin.[5] The HMB-45 stains are considered to show greater specificity for melanoma than S-100 protein stains.[5] The immunoperoxidase stains in our patient's case showed positive findings in S-100 protein and HMB-45 stains.

However, these findings may be inconsistent, and the diagnosis of a primary oral mucosal melanoma requires the careful search for and the exclusion of any other suspicious cutaneous or mucosal lesions.[4]

In our patient's case, there was no history of melanoma-like lesions and no suspicious cutaneous or mucocutaneous discolorations or masses detected by examination of her chest, abdomen, extremities and head or neck, including the nasal cavity, pharynx and larynx. Hence, by correlating both physical and histopathology findings, we confirmed the diagnosis of primary melanoma.

Surgery is believed to be the most effective treatment for melanoma.[1] Wide resection with a surgical margin of 2–5 cm is necessary for cutaneous melanoma but is difficult to achieve in its oral form owing to evident anatomic restrictions. The role of radiotherapy is controversial. Many authors believe melanoma to be a radioresistant neoplasm, and hence, radiotherapy is frequently used in palliative therapy. However, its adjunctive role with chemotherapy has shown effectiveness in the primary management of unresectable tumours. In our patient's case, taking into account the free surgical margins of the resected tumour, she did not receive any adjuvant chemo- or radiotherapy.

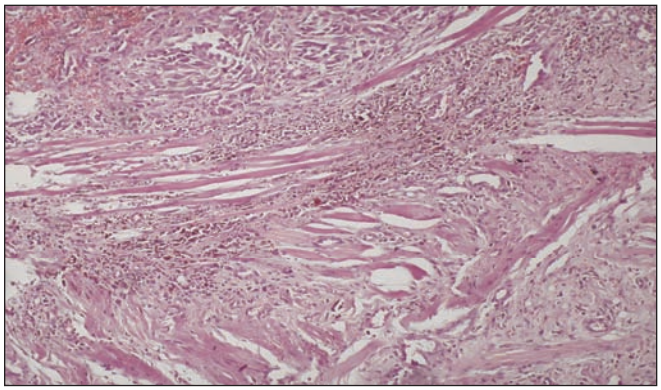

Fig. 3. Malignant melanoma of tongue. This field showed sheets of spindle-shaped malignant melanocytes infiltrating the superficial tongue muscles. Gross depth of infiltration was 0.5–1.0 cm (hematoxylin and eosin stain, original magnification ×100).

In general, the prognosis for patients with oral malignant melanoma is worse than for patients with cutaneous lesions. The 5-year survival rates are 6.6%–20.0%.[3] Several factors may contribute to this poor prognosis, including lack of symptoms early in the disease, difficulty in achieving wide radical excision because of anatomic limitations and rich blood supply that may facilitate heamatogenous spread.[1]

The prognosis for patients with oral malignant melanoma is poor, with a 5-year survival rate between 11.0% and 18.0%. Late diagnosis often coincides with an extensive metastatic tumour. After surgical ablation, recurrence and metastasis are frequent events, and most patients die of the disease in 2 years. A review of the literature indicates that the 5-year survival rate is within a broad range of 4.5%–48.0%, but a large cluster occurs at 10.0%–25.0%. Early diagnosis should be promoted by careful oral examination and early biopsy of pigmented and nonpigmented suspicious lesions to improve the prognosis of patients with oral malignant melanoma.

Competing interests: None declared.

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