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Recurrent Oral Melanoma: A Case Report and Challenges in Management in Poor Resource Economy

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Abstract Oral melanoma is a rare aggressive neoplasm which accounts for only 0.2-8% of all reported malignant melanomas. The common sites of its occurrence are the palate and gingiva with the maxillary arch being affected 80% of the time. Because of their presence at relatively obscure areas in the oral cavity, most of the malignant melanomas of the oral cavity are diagnosed at a late stage. We report a case of oral melanoma in a 55-year-old post-menopausal woman who had surgical excision followed by chemo-radiation but suffered recurrence. The challenges of management and the poor prognosis of the case are emphasized in this case report.

Keywords: *oral melanoma, recurrence, palate, gingiva, management challenges*

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1. Introduction

Malignant melanoma is a neoplastic proliferation of melanocytes derived from neural crest which can arise from a benign melanoctytic lesion or de novo from melanocytes within otherwise normal skin or mucosa [1-5]. Oral melanoma affects the mucus membrane of the oral cavity and can be primary or metastatic (secondary).

It is extremely rare accounting for only 0.2 – 8 %[6] of all malignant melanomas although higher incidence has been reported in India, Africa, and most especially Japanese population [7,8]. Oral Melanoma accounts for 1.6% of all head and neck malignancies [9]. It is considered to be the most deadly and biologically unpredictable of all human neoplasms, having the worst prognosis [1,9]. The common sites of occurrence are the palate and gingiva with the maxillary arch being affected 80% of the time [1]. Because of their presence at relatively obscure areas in the oral cavity, most of the malignant melanomas of the oral cavity are diagnosed at a late stage [10].

The age range of patients with oral melanoma is from 35 to 80 years [11,12]. The main aim of the article is therefore to report a case of recurrent oral melanoma in a 55 year old Nigerian woman and highlight the challenges of management of this condition in a resource poor setting.

2. Case Report

A 55-year-old Nigerian woman presented to our clinic with a 2-month history of painless pigmented mass of the gingivae. She volunteered a history of topical application of herbal concoction to the swelling with associated history of spontaneous bleeding. Her medical history was remarkable for recent diagnosis of hypertensive heart disease which was well controlled. Clinical examination revealed a firm, localized swelling, blue- black in color in relation to the upper left lateral incisor (22) and upper left second premolar (25) (Figure 1). The swelling is sessile, not tender with irregular margins. She had complete set of permanent dentition of which all surrounding teeth are firm. Neck palpation revealed no abnormal findings and there was no pigmented skin lesion. Complete blood count, biochemical profile, urinalysis, peri apical and chest X-ray examination yielded no abnormal findings.

Figure 1. shows a firm, irregular, blue- black swelling, in affecting the upper left lateral incisor (22) and upper left second premolar (25)

Surgical excision of the lesion was carried out via gingival margin and vestibular incisions made along lesional perimeter. The underlying bone was curetted under copious 0.9% normal saline irrigation after which the exposed bone was allowed to granulate normally.

Grossly we received an irregularly shaped almost entirely black-brown tissue that weighed 10 grams and measured 3cm x 3cm x 3.5cm. It was firm in consistency. Serial sections showed brown-black surfaces. It was partly embedded in five blocks.

Microscopically, the lesion consisted of melanocytic cells which are oval to spindle with vesicular to hyperchromatic and pleomorphic nuclei disposed in sweeping fascicles (Figure 2 & Figure 3). The tumour cells are diffusely distributed in the stroma and are laddened with melanin pigments, accompanied by abnormal mitotic figures exceeding 5/10HPF consistent with a diagnosis of pigmented malignant melanoma.

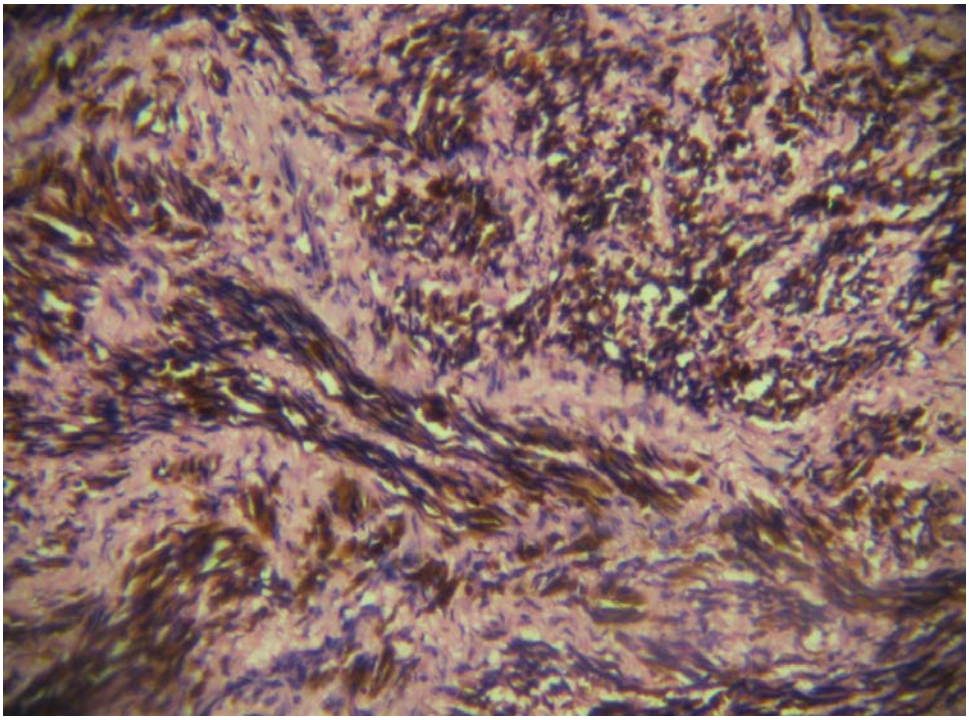

Figure 2. Photomicrograph showing melanocytic cells which are oval to spindle with pleomorphic vesicular to hyperchromatic nuclei and abundant melanin pigments

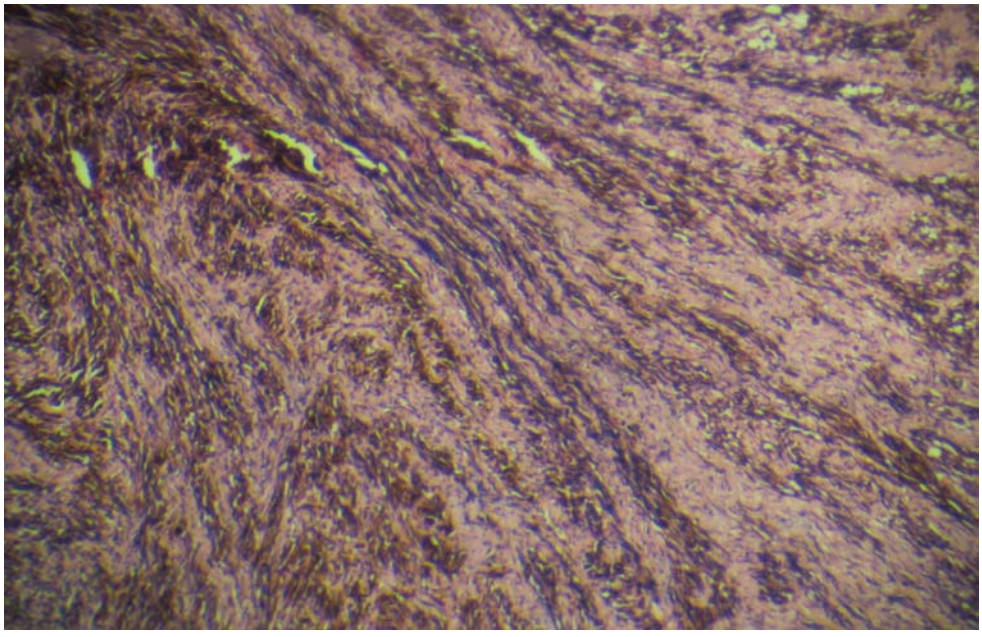

Figure 3. shows photomicrograph of the same lesion showing melanocytic cells in sweeping fascicles

Work-up for distant metastases with computed tomography (CT) was planned but patient declined due to financial constraint. A month after initial surgical intervention, she suffered recurrence. The patient was counseled for radio- chemotherapy which she received 3 months after initial surgery. She could not however complete the sessions because of high cost of chemotherapeutics agent.

3. Discussion

Malignant melanoma of the oral cavity is extremely rare. The rarity of the lesion is corroborated by the preponderance of isolated case reports in the literature [6,7,13]. Soyemi et al' [14] retrospective study of 111 cases of oro-pharyngeal tumours seen in three General Hospitals in Lagos, Nigeria from January 2003 to December 2007 revealed no case of mucosal melanoma. Likewise, a search of register of maxillofacial pathologies in our centre from January 2008 to date revealed the index case as the only case diagnosed.

Eighty percent of cases involve the gingival and palate while the mandible, buccal mucosa, lips, tongue and floor of mouth are affected to a lesser extent. The present case also highlights the gingiva as a common location. The reason for this is not very clear however it may be related to the relative high melanin concentration in the gingivae [1].

The patient gave a history of repeated topical application of herbal concoction to the lesion which may be a predisposing factor. Local herbal concoctions are frequently used in African population because of beliefs in their efficacy. The contents of these concoctions though not clearly known frequently contain caustic agents injurious to the mucosa. None of the other risk factors associated with malignant melanoma such as Ultra Violet radiation, PUVA therapy, immune-suppression, denture irritation, exposure to tobacco, chemicals, petroleum, and printing products was present in the patient. Higher prevalence of oral pigmented lesions has been previously demonstrated among smokers thus suggesting cigarette smoking as a risk factor. [15] Conversely, the history of herbal application may also be a cause of misdiagnosis as the clinical significance of the black discoloration may easily be mistaken for the discoloration caused by the concoction. Other oral pigmented lesions such as melanotic pigmentations, amalgam tattoo, melanoacanthoma, melanotic macule, oral nevi, racial pigmentation, smoking-associated melanosis, post-inflammatory pigmentation, medication melanosis may mimic oral melanoma especially at early stages. [7] Although most pigmented lesions in the oral cavity are benign and of no clinical consequence, [1] clinicians must maintain a high index of suspicion for oral lesion with brown-black discolorations with more sinister biologic behaviour. This serves to emphasize the importance of histopathological examination of all pigmented oral specimen.

Oral melanoma is a biologically aggressive neoplasm with a poorer prognosis than its cutaneous variant [7,12]. The main prognostic factor appears to be lymph node compromise. There was no regional lymph node enlargement at presentation in the present case, although up to 50% of cases have been reported to have lymphadenopathy at presentation [16], the use of more advanced imaging techniques like CT and MRI which have unique characteristics to delineate and detect occult lymph node metastasis may be useful [17]. However, the cost of these advanced imaging techniques is not readily affordable in third world countries particularly in our centre which is semi -urban. These challenges are well known and have been variously reported in the management of the lesion in resource poor countries of Africa [4].

Greene *et al*. [18] proposed the criteria for diagnosis of primary oral malignant melanoma which include demonstration of melanin in oral mucosa, presence of junctional activity and inability to demonstrate extra oral primary melanoma. The present case fulfilled the above criteria. Furthermore, Meleti et al [19] proposed a histological classification of oral melanoma based on microscopic pattern which classifies lesion into melanoma in situ, invasive, and combined. Our patient's lesion displayed an invasive form although it remains to be seen of what prognostic importance this is. The present case also showed relatively rapid progression with early bone destruction and high recurrence rate. This is in spite of the prompt referral for post operative chemo radiotherapy. It must however be noted that the delay in commencement of radio chemotherapy and sub optimal chemo-radiotherapy regimen received by the patient due to financial and logistic reason may be a factor responsible for the rapid progression.

Most authors [20,21] agree that excisional biopsy with a wide margin followed by adjuvant chemo radiation offers the best treatment for oral melanoma. Early wide surgical excision was carried out for our patient however she could not receive optimal post operative chemo radiation due to financial and logistic reason. This may be responsible for the early recurrence seen in our patient.

In conclusion, a high index of suspicion especially for pigmented oral lesion may aid early detection which can considerably improve prognosis and reduce the burden of cost of treatment in our setting. The role of advanced imaging techniques for evaluation and treatment monitoring cannot be over-emphasized.

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