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Spindle Cell Melanoma with an Insight and Review of Literature - Case Report

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Case Report

History

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ABSTRACT

Spindle cell melanoma is a rare histological variant of malignant melanoma that is potentially aggressive, most often arising on the skin but only rarely in the oral cavity. Its varied clinical manifestations and resemblance to other spindle cell or amelanotic lesions make diagnosis particularly challenging for dental practitioners. Prognosis depends on tumor size, metastatic spread, and advanced diagnostic investigations. The aim of this report is to present and analyze the case of a middle-aged female diagnosed with oral spindle cell melanoma, highlighting its distinctive histopathological features, the diagnostic difficulties encountered, and the therapeutic approaches considered, thereby emphasizing the importance of early recognition and comprehensive evaluation in improving patient outcomes.

Keywords: Mucosal melanoma, malignant melanoma, Oral melanoma, spindle cell melanoma, spindle cell variant

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Introduction

Melanoma is a malignant tumor of melanocytes derived from neural crest cells, most commonly of cutaneous origin. Elder et al. modified the WHO classification of melanoma into three categories: melanoma associated with cumulative solar damage (CSD), melanoma not associated with CSD, and nodular melanoma, each reflecting distinct mutagenic pathways in melanoma development.[1]

Mucosal melanoma (MM), a subtype not associated with CSD, arises from mucosal linings, with the head and neck region being the most commonly affected site.[2] Although its exact etiology remains unclear, potential risk factors include alcohol consumption, cigarette smoking, and formaldehyde exposure.[3,4] MM is rare in the oral cavity; when present, it most frequently involves the palate, followed by the maxillary gingiva.[5]

Spindle cell melanoma (SCM) is a rare histological variant of malignant melanoma. It often mimics amelanotic or reactive lesions and may occur anywhere in the body, posing a considerable diagnostic challenge.[6] Because of these varied clinical presentations; oral melanoma remains a significant diagnostic dilemma for dental practitioners, as also highlighted in recent case reports.[7]

Herein, we present a rare case of spindle cell melanoma of the anterior maxillary gingiva, discuss its diagnostic complexity, and review relevant literature.

Case Report

A 44-year-old female patient was referred to the Department of Oral Medicine and Radiology from a rural hospital with a chief complaint of swelling in the upper front teeth region since two months. The swelling was slow in progression, asymptomatic, and associated only with mild difficulty in chewing. The patient reported that the swelling had appeared following extraction of teeth 12 and 13. She had no relevant medical or family history and denied the use of tobacco, alcohol, or other deleterious habits.

Extraoral examination revealed mild swelling at the philtrum with loss of the right nasolabial fold. Intraoral examination showed a sessile, lobulated swelling measuring approximately 3 × 2 cm, extending from the distal of 11 to 14, obliterating the vestibular space (Figure 1). The swelling was well-defined, pink-red, firm, and non-tender, with mild diffuse melanin pigmentation over the labial and palatal mucosa of 11. Mobility was noted in teeth 11, 14, and 15. A provisional diagnosis of fibroma was given.

Orthopantomogram (OPG) revealed complete loss of bone extending from the mesial aspect of 11 to the distal of 15, reaching the floor of the right maxillary sinus (Figure 2). An incisional biopsy was initially performed, considering the clinical appearance of a benign lesion. Histopathology revealed granulation tissue with proliferating blood vessels lined by plump endothelium

and intervening lymphomononuclear infiltrates, suggestive of chronic nonspecific inflammation with granulation tissue. Since these findings did not correlate with the clinical picture, advanced imaging was advised

Cone-beam computed tomography (CBCT; CS 9600 software, FOV: 6 × 6 cm) revealed a well-defined radiolucency measuring about 26.7 × 13.3 mm on the axial section and 14.3 mm on the coronal section. The lesion extended anteroposteriorly from 11 to the distal aspect of 15 and superoinferiorly from the floor of the maxillary sinus to the alveolar crest. Extensive destruction of the labial, buccal, and palatal cortical plates and the floor of the right maxillary sinus were noted (Figure 3). These findings raised strong suspicion of malignancy.

Ultrasonography of the neck was performed to evaluate possible cervical nodal involvement. No evidence of metastatic lymphadenopathy was detected. Ultrasonography was chosen due to its non-invasive, cost-effective nature and reliable diagnostic value in assessing lymph node morphology in resource-limited rural settings. Advanced modalities such as PET-CT or MRI could not be utilized due to financial and accessibility constraints.

Considering the radiographic evidence of malignancy, the patient underwent a right maxillectomy. The resected specimen (Figure 4) showed a tumor composed of nests, fascicles, and clusters of atypical cells infiltrating the connective tissue and bone. The tumor cells displayed elongated or oval hyperchromatic nuclei, prominent nucleoli, and moderate amounts of eosinophilic cytoplasm. Occasional multinucleated tumor giant cells, areas of lymphoplasmacytic infiltration, and focal melanin pigmentation were also noted (Figure 5).

To establish a definitive diagnosis, an immunohistochemical panel was employed. The tumor cells were diffusely positive for Vimentin, S-100 protein, and SOX-10, while negative for cytokeratin (CK), p63, Melan-A, HMB-45, and MiTF. Retained H3K27ME3 expression further supported the diagnosis of spindle cell melanoma.

The patient was placed on a structured follow-up protocol with monthly clinical examinations for the first six months, followed by three-monthly reviews. Each visit included intraoral and extraoral examination, nodal assessment, and radiographic evaluation. At six, nine, and twelve months, the patient showed satisfactory healing without evidence of recurrence or metastasis. At the time of reporting, the patient remained disease-free under regular surveillance.



Figure 1. Intraoral photograph showing spindle cell melanoma of the maxillary gingiva.

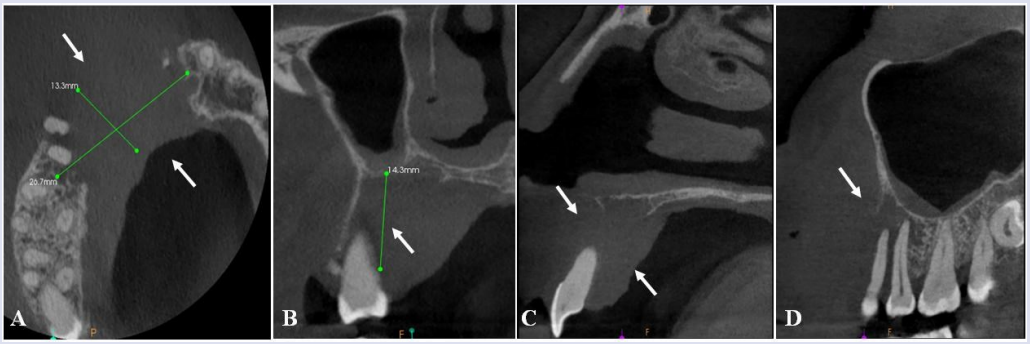


Figure 2. Orthopantomogram (OPG) showing bone destruction in the anterior maxilla.

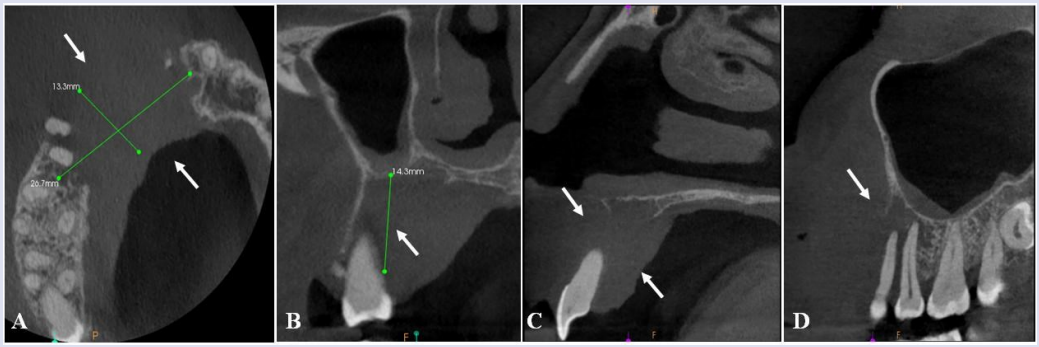


Figure 3. Cone-beam computed tomography (CBCT) views of the maxillary lesion. (A) Axial view showing a well-defined osteolytic lesion with measured dimensions of 26.7 mm × 13.3 mm (green lines) and cortical perforation of the labial and palatal plates (arrows). (B) Coronal view demonstrating the vertical extent of the lesion (14.3 mm) extending from the alveolar crest to the floor of the maxillary sinus (arrow). (C) Sagittal view depicting cortical erosion and superior extension towards the maxillary sinus floor (arrows). (D) Sagittal slice highlighting aggressive resorption of the anterior maxillary alveolar bone and sinus wall involvement (arrow).

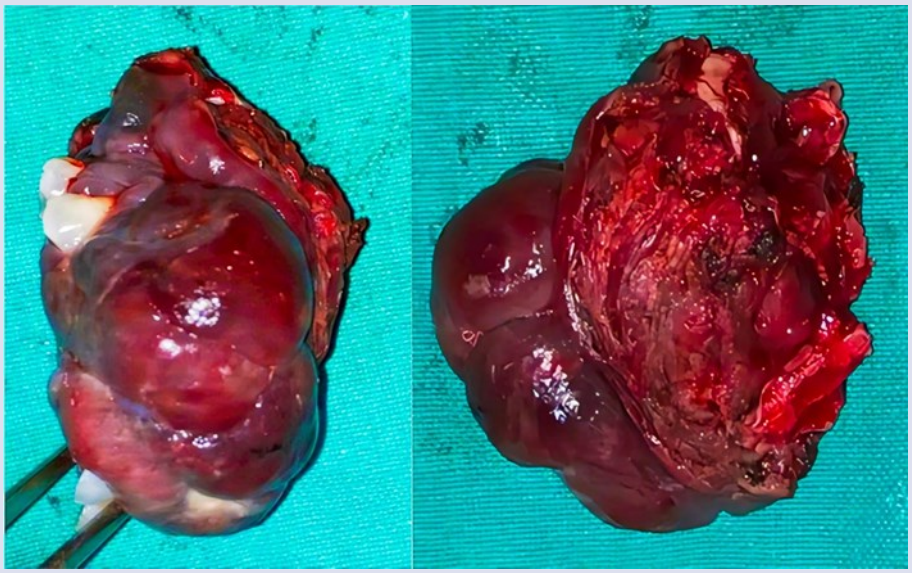


Figure 4. Surgical specimen following right maxillectomy.

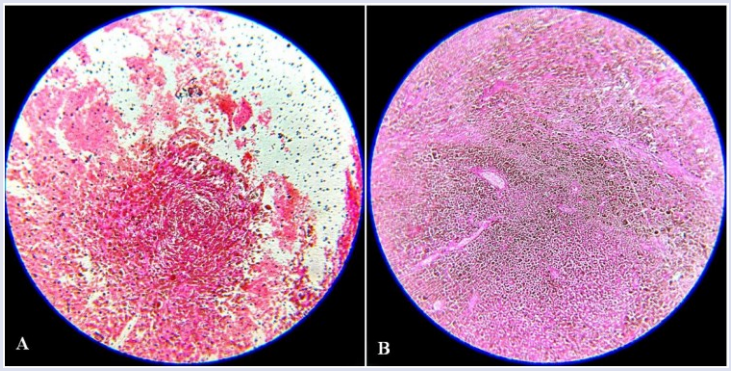


Figure 5. Histopathological sections showing spindle-shaped tumor cells with focal melanin pigment deposition. (A) Section showing oval elongated nuclei, conspicuous nucleoli, and moderate cytoplasm (H&E, ×10).(B) Section showing inflammatory infiltrate comprising lymphocytes, plasma cells, and melanin pigmentation in tumor cells (H&E, ×10).

Discussion

Oral mucosal melanoma (OMM) is a rare but aggressive malignancy, accounting for approximately 0.2–8% of all melanomas,[8] with the hard palate and maxillary gingiva being the most frequently affected intraoral sites.[9,10] Our patient's lesion in the anterior maxillary gingiva is consistent with this distribution, although the clinical presentation was atypical. Spindle cell melanoma, a histological subtype, comprises roughly 3–14% of all melanomas and frequently poses a misdiagnosis due to its resemblance to other spindle cell neoplasms.[6]

Most oral melanomas are pigmented, but up to 20% lack pigmentation and may appear as pink or red swellings that resemble benign lesions.[3,11,12] In our case, the lesion was predominantly amelanotic with only faint pigmentation, explaining the initial clinical impression of fibroma. Similar diagnostic challenges have been described by Tanaka et al. in their investigation of amelanotic oral melanomas.[11] Histopathologically, small incisional biopsies may yield inconclusive findings in spindle cell–dominant melanomas. In our patient, the initial biopsy revealed only nonspecific granulation tissue, a limitation also highlighted by de Andrade et al., who reported that repeat or excisional biopsies were often required to establish a definitive diagnosis.[12] This emphasizes the importance of integrating clinical, radiographic, and histopathological features for accurate diagnosis.

Our CBCT revealed aggressive cortical destruction and sinus floor involvement, which prompted definitive surgical intervention. Comparable radiographic features have been reported in oral mucosal melanoma cases, where coronal CT scans demonstrated bone erosion extending to the maxillary sinus floor.[13] Ohnishi et al. also observed that maxillary melanomas frequently exhibit aggressive bone invasion on imaging, even in the early stages.[14]

Histologically, oral melanomas can exhibit epithelioid, spindle, plasmacytoid, or mixed cell populations.[12] In our case, spindle cell morphology predominated, making differentiation from other spindle cell tumors such as sarcomas, spindle cell carcinoma, and reactive lesions critical. Immunohistochemistry (IHC) proved decisive. Although conventional melanocytic markers such as HMB-45 and Melan-A were negative, the tumor was diffusely positive for S-100 and SOX-10, with retained H3K27ME3 expression. This immunoprofile has been observed in spindle-rich or desmoplastic melanomas and is consistent with findings by de Andrade et al. and Ohnishi et al., who reported S-100 and SOX-10 as more reliable markers in mucosal melanomas compared to HMB-45 or Melan-A. Our case therefore reinforces the need for a broad immunohistochemical panel when evaluating spindle cell lesions of the oral cavity.[12,14]

Consistent with current recommendations, we performed a wide surgical resection (right maxillectomy). The short-term postoperative course has been favorable, yet vigilant long-term surveillance remains crucial given the historically poor prognosis of OMM. Turkish reports support this: a maxillary-gingival melanoma case documented low five-year survival and frequent late metastases, reinforcing the need for prolonged follow-up.[15] Similarly, a Turkish systematic review and case report on oral mucosal melanoma in situ emphasize the necessity of long-term monitoring, even when diagnosed at an early stage.[16]

Conclusions

Primary oral melanomas are rare and often pose diagnostic challenges, especially when amelanotic and clinically resemble benign lesions. In this case, the incisional biopsy was inconclusive, and the final diagnosis of spindle cell melanoma was achieved only after a broad immunohistochemical panel, with SOX-10 and S-100 proving decisive. This emphasizes the importance of comprehensive diagnostic workup and structured follow-up for timely management and improved prognosis.

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None.

Conflicts of Interest Statement

No Conflicts of Interest.

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