CASE REPORT

An unusual variant of primary oral malignant myxoid melanoma with varied clinical significance

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ABSTRACT

Malignant myxoid melanoma histologically presents as atypical spindle cells in a background of dense mucin deposition in connective tissue. It a is a rare variant of malignant melanoma. A substantial overlap in clinical and pathologic features of this variant with other arrays of myxoid containing tumors makes a definitive diagnosis challenging. The myxoid component seen is a response to the tumor cells and therefore is usually seen in metastatic lesions. Though few cases have been reported on skin and face, this case reports the first case of primary myxoid malignant melanoma in the oral cavity. To conclude, proper co-relation of histopathological and immunohistochemical study is the gateway for final diagnosis of myxoid variant of malignant melanoma.

Keywords

Immunohistochemistry, Malignant Melanoma, Melanocytes, Myxoid, Spindle cells

INTRODUCTION

Malignant Melanoma is a rare tumor arising from melanocytes derived from neural crest cells or their precursors and accounts for 0.5 % of oral neoplasm and 1-2 % of all melanomas. The etiopathology of such lesions is still ambiguous although sun exposed areas, tobacco smoking, regular alcohol usage, and HPV contamination has been suggested as probable risk factors.(1,2) Oral melanoma occurs between 55 to 66 years of age with no gender predilection.(1,3)

Besides the usual clinical variants, atypical metaplastic change in the stroma of melanoma may result in various unusual histologic variants such as small cell melanoma, balloon cell melanoma, myxoid, osteoid, signet-ring cell, fibroblastic, desmoplastic, condroid, and rhabdoid melanoma which have been reported on skin [4]. Though myxoid variants has been reported in previous literatures on skin, this case is the first case of myxoid metaplasia in malignant melanoma located on alveolar mucosa.

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Case History

A female aged 98 years visited the Department of Oral medicine and Radiology, Kalinga Institute of Dental Sciences, Bhubaneswar, Odisha with a swelling in upper left posterior region with a duration of 6 months. The swelling was of gradual onset, non-tender, rapidly progressive hindering mouth closure and was associated with mild pain in the later stage.

Extraorally, there was a diffused swelling of about 6cm x 4cm in the greatest dimension in left maxillary area (Figure 1a). Intraorally, a large exophytic growth was seen involving the left maxillary gingiva, alveolar mucosa and palatal mucosa extending from the midline till the maxillary tuberosity measuring approximately 7cm x 5cm was seen which hindered the mouth closure. The surface was smooth and partly pigmented. The teeth seemed to be displaced. (Figure 1b) The lesion was soft in consistency, fixed to the underlying structures, tender on palpation, and bleeding on provocation was present. The second premolar of the affected side was grade II mobile.

The Cone Beam Tomography showed a lesion of 7.2 X 4.6 cm extending from the left maxillary incisors to the molar region and maxillary tuberosity. The edges of the left maxillary alveolar bone were ragged. Horizontal and vertical bone loss with loss of lamina dura was seen. The left second premolar had a floating appearance. (Figure 2) An incisional biopsy was done and tissue was sent to the Department of Oral and Maxillofacial Pathology for histopathological confirmation.

Initial histopathology examination of incisional specimen showed epitheloid cells containing abundant melanin pigments juxta epithelially. Complete surgical excision of the lesion with 5 mm safety margin was done and the mass was sent for histopathologic confirmation and immunohistochemical analysis. Gross features showed a partly pigmented tan brown mass whose cut surface had a slimy consistency.

On histopathology of the excised specimen, an atropic thinned out stratified squamous epithelium covering a myxomatous connective tissue component was noticed. One part of the tissue section showed highly cellular area consisting of spindle cells showing malignant features like cellular atypia, aberrant nuclear morphology and anisocytosis. The connective tissue component shows predominantly myxoid areas (Figure 3). Tumor areas showed a strong positivity for S-100 protein, vimentin, Melan A and HMB45A (Figure 4). Alcian blue at low PH showed positivity for myxoid areas. Routine haematological and biochemical investigations including PET-CT scan was done to rule out metastasis. The case was diagnosed as primary myxoid malignant melanoma. A regular 6 monthly follow up was advised.

DISCUSSION

The malignant melanoma of the oral cavity account for only 0.5% of melanomas.. Various variants in this malignant neoplasm of melanocytes have been encountered such as, spindle cell melanoma, neurotropic melanoma, osteoid producing melanoma and desmoplastic melanoma among others. Myxoid melanoma is one such rare entity which has been reported on skin and face [4]. The present case is the first one to be reported in an unusual site of left maxillary alveolar mucosa with a rare histopathological finding a 98 year old female.

Melanomas have a male predominance. Majority of the reports stated an average age of 60 years and 54 years for primary and metastatic myxoid melanoma respectively [5]. The present case was a female who was 98 year old with primary tumour.

A delayed diagnosis is often a result of its painless nature leading to later symptoms like bleeding ulceration or increase in size [1]. Pain is not a manifestation in melanoma as in our case which had caused a delay in approach for treatment.

A “ABCD” differentiating criteria [6] for malignant melanoma to characterize from other benign pigmented lesions was used for arriving at a preliminary diagnosis malignant melanoma. The present case met all these criterias of asymmetry, border irregularity and 0.6 mm or greater diameter.

On histopathology, this case showed highly cellular area consisting of aberrant spindle cells admist the predominantly myxoid areas in the connective tissue. Myxoid component can be appreciated in arrays of lesion such as myxoid malignant fibrous histiocytoma, malignant melanotic schwannoma, desmoplastic melanomas, myxoid chondrosarcoma and myxoid rhabdomyosarcoma [4]. The abundant myxoid tissue consisting of gelatinous mucopolysaccharide matrix of sulfated and non sulfated glycosaminoglycans in the

extracellular matrix are charactistics of these lesion.[7] Proper clinical understanding, histologic findings and immunochemistry can lead to correct diagnosis. In our case the myxoid stroma was positive for mesenchymal acid mucopolysaccaride like Alcian blue at low PH and negative for neutral mucins like mucicarmin and PAS diastase. Also, absence of the mucin like material within the cytoplasm of the tumor cells was noticed. Therefore, it was inferred that the myxoid material is not produced by the tumor cells but might be the stromal cells' response to the tumor. Literatures suggest a meticulous evaluation for primary tumor in such variant since they commonly constitute of metastatic tumor tissue [8]. So, a prompt PET-CT scan among other routine haematological and biochemical investigations was done to rule out metastasis. Thus, an apt diagnosis of primary oral myxoid malignant melanoma was reached.

A rational explanation of myxoid melanomas to be often confused with other mucin-containing neoplasms might be that myxoid areas have been rarely reported in malignant melanoma. The myxiod component is actively secreted by neoplastic cells and is not a normal tissue in adults. [7] Patel et al. [5] found higher number of mast cells and hypothesized that they in association with transforming growth factor beta might be responsible for stimulation of mucin secretion by fibroblast, which also aids the invasiveness of the tumor. This might explain the large tumor size of 7cm x 5cm in the present case.

Generally, wide surgical excision with adequate tumor free margins is the treatment of choice for malignant melanoma. It may be with or without neck dissection[9]. A localized form of oral melanoma can be controlled by radiotherapy, as opposed to skin melanoma. Distant metastases can be prevented by chemotherapy and immunotherapy. Oral melanomas have a five-year survival rate of 4.5% to 29%, with a median survival rate of 18.5 months after initial diagnosis [10]. Wide surgical excision was performed in the present case. Owing to her age radiotherapy was not advised. The patient is tumor free since 24 months.

CONCLUSION

Most oral melanomas are large at presentation and have a poor prognosis in comparison to skin melanomas. The nature of myxoid tumors being more aggressive and with a higher metastatic potential make it a mandate for a definitive prompt diagnosis. The painless nature of this lesion and a delay in diagnosis due to such unusual histologic finding may pose as a major diagnostic pitfall and lead to poor prognosis. Due to its aggressive nature and tendency to metastasize they should be addressed with utmost care & vigilance.

Figure 1- (A) Extra oral View (B) Intra-oral View

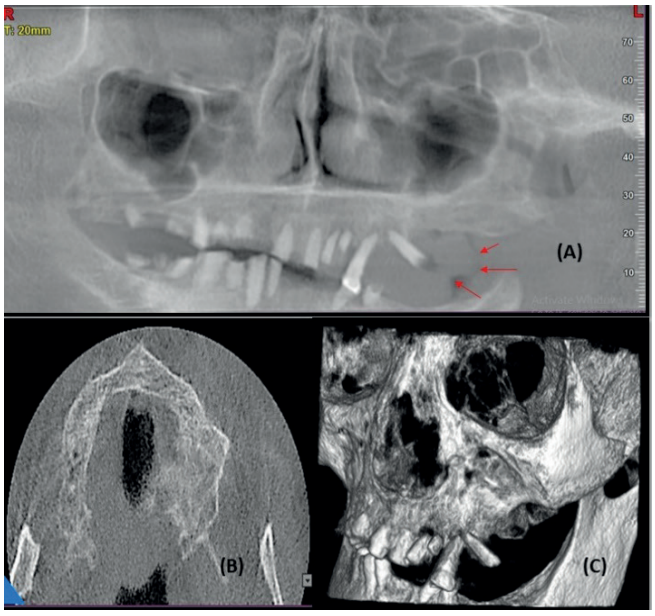

Figure 2: Radiovisiography showing Radiolucency in the maxilla

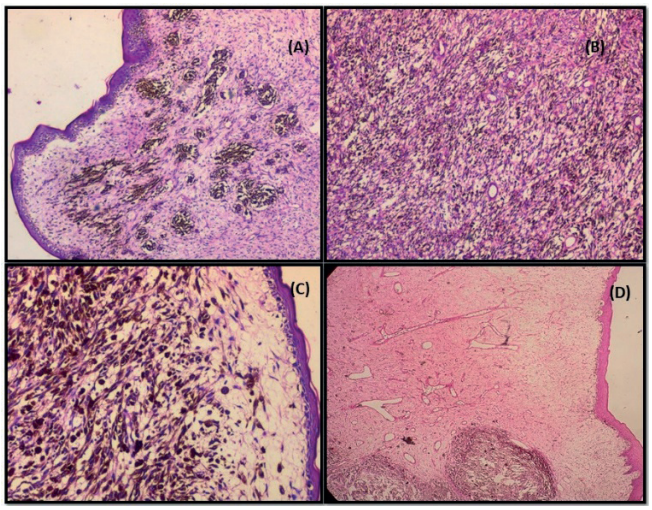

Figure 3: (A) Histopathology showing an atropic thinned out stratified squamous epithelium covering a myxomatous connective tissue component. (B) (C) (D) Highly cellular area consisting of spindle cells showing malignant features like cellular atypia, aberrant nuclear morphology and anisocytosis.

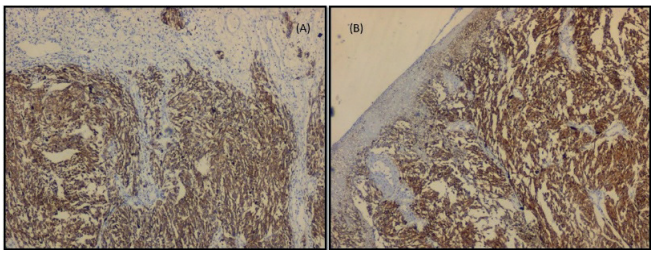

Figure 4: Tumor areas showed a strong positivity for Melan A and HMB45A.

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