International Journal of Otorhinolaryngology and Head and Neck Surgery
Sharma J et al. Int J Otorhinolaryngol Head Neck Surg. 2021 May;7(5):896-899
http://www.ijorl.com
pISSN 2454-5929 | eISSN 2454-5937

DOI: https://dx.doi.org/10.18203/issn.2454-5929.ijohns20211591

Case Report

Malignant melanoma oral cavity: case report of two patients from sub-Himalayan region

Jyoti Sharma[1]*, Manish Gupta[1], Amit Saini[2]

[1]Department of Radiotherapy, Tertiary Cancer Centre, IGMC Shimla, Himachal Pradesh, India
[2]Otolaryngology, Head and Neck Surgery, Dr. Radhakrishnan Government Medical College, Hamirpur, Himachal Pradesh, India

Received: 14 March 2021
Revised: 18 April 2021
Accepted: 19 April 2021

***Correspondence:**
Dr. Jyoti Sharma,
E-mail: jyotisharma217@gmail.com

ABSTRACT

Malignant melanoma of the oral cavity is an exceedingly rare tumor representing 0.2 to 8% of all melanomas. Mucosal melanomas are extremely rare and aggressive neoplasms. Patient reporting to the clinician with a pigmented lesion should raise suspicion in the first visit itself and should be further investigated so as to detect this dreaded malignancy at an earlier stage and thus managed appropriately. We present two such rare cases who reported at our centre treated with different treatment modalities and had different responses to treatment. In first case report 65 year old male patient diagnosed with malignant melanoma of left upper alveolus underwent multiple modalities of treatment like surgery, chemotherapy, radiation therapy but unfortunately as he defaulted post-surgery and also due to COVID-19 lockdown restrictions he was treated in various centres and finally the result was inoperable residual gigantic mass resistant to chemotherapy and radiation therapy. In second case report, 82 year old male patient reported with malignant melanoma hard palate having good response to initial chemotherapy. He was planned on hypo-fractionated radiotherapy in view of his old age but he refused radiation treatment and is on oral temozolamide and thalidomide with stable disease and good quality of life since past 6 months.

Keywords: Mucosal melanoma, Oral cavity, Radiotherapy, Targeted therapy

INTRODUCTION

Malignant melanoma of oral cavity i.e., mucosal melanomas are rare neoplasms originating from the malignant transformation of the melanocytes. These cells are found in the basal layer of the oral mucosal membrane and are thought to be of neural crest origin.[1,2] A definitive precursor lesion for mucosal melanoma has not been known but Hicks MJ et al have mentioned that a atypical melanocytic hyperplasia may represent a proliferative phase before overt tumorigenesis occurs in mucosal melanoma. In contrast to the cutaneous melanoma whose etiological factors have been described as sun exposure, risk factors of mucosal melanomas are unknown. No definitive evidence suggests any relationship to smoking, alcohol and ill-fitted dentures to which mucosa is constantly exposed.

Mucosal melanoma of the oral cavity is a rare tumor with incidence of 0.2 per million.[3] The common sites of its occurrence are the palate and gingiva with the maxillary arch being affected 80% of the time.[4]

Surgical resection of the lesion with negative margins is the treatment of choice which offers chances of cure. There is role of chemotherapy and radiotherapy in

adjuvant and palliative setting. Here we present two cases of malignant melanoma of oral cavity who presented in our centre with different results of treatment.

CASE REPORT

Case 1

65 year old male patient was registered with chief complaint of large, ulcerated mass in oral cavity since past 2 years. There was history of bleeding from the mass.

He was a diagnosed case of malignant melanoma left upper alveolus according to the records available and he was seeking treatment at higher centre (1st centre). As per records initial CECT revealed heterogeneously enhancing mass involving left upper alveolus gingival mucosa. Metastatic workup was done with the help of PET scan which revealed hypermetabolic soft tissue density mass in left vestibule of mouth with its epicentre in left upper gingiva.

He underwent left upper alveolectomy along with MND III+skin grafting and reconstruction. Detailed HPE was suggestive of poorly differentiated malignant tumor with no lymphovascular space invasion, no perineural invasion, all margins free, all 30 lymph nodes free from tumor.

Unfortunately, patient lost to follow up for 10 months and received no adjuvant treatment.

When he reported at the same centre, PET scan was done to know the status of the disease which revealed metabolically active soft tissue thickening in left upper alveolar region with right pulmonary nodule. Punch biopsy from left upper alveolus revealed malignant melanoma. IHC was positive for S100, HMB45, melan A. Patient was put on injection nivolumab based immunotherapy. He received two cycles of the same immunotherapy but was not able to report to higher centre due to COVID-19 lockdown.

He had to shift to nearby cancer centre (2nd centre) where he received two cycles of cisplatin and dacarbazine based chemotherapy.

There was disease progression and again shifted to our centre (3rd centre). His chief complaints when he presented at our centre were bleeding from the mass since past 20 days, increase in the size of the mass, headache and dizziness since past 20 days. There were no co-morbidities. In general physical examination pallor was present. Systemic examination was with in normal limits. Local examination revealed large 10×8 cm ulceroproliferative growth involving left sided upper alveolus, buccal mucosa with restricted mouth opening and tongue protrusion. Posterior extent could not be assessed. (Figure 1a) In his routine investigation pallor was present. Rest of the examinations were with in normal limits. He was given palliative radiotherapy single fraction of 6 Gy by direct anterior field. Patient reported no response to Rx and was started on palliative chemotherapy based on paclitaxel and carboplatin. There was no response to chemotherapy was well. He was considered for change in chemotherapy and was started on oral temozolamide. Despite giving 3 cycles of oral chemotherapy there was significant progression in size of lesion. Result was gigantic mass obliterating almost entire oral cavity (Figure 1b). He was given palliative radiation therapy 6 Gy single fraction. prognosis was explained and patient was sent home for best supportive care.

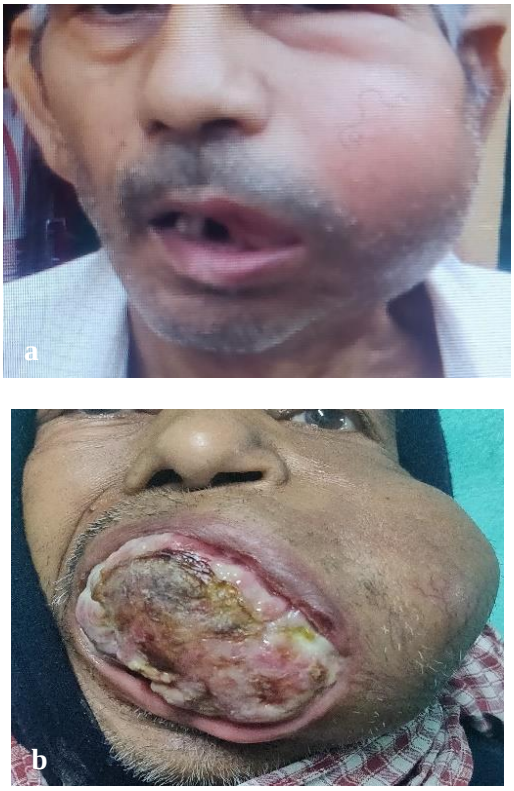

Figure 1: (a) Initial presentation of case 1 at registration, (b) case 1 shows gigantic progression despite surgery chemotherapy and radiation; patient defaulted for 10 months post-surgery; he had delays in treatment owing to COVID-19 lockdown restrictions.

Case 2

82 year old male patient presented with chief complaint of pain in mouth and black-coloured mass over hard palate on left side with off and on bleeding. He initially presented in department of oral and maxillofacial surgery where he was investigated and biopsy was done revealing malignant melanoma. He had no co-morbidities. There was history of smoking for past 30 years. There was history of alcohol intake for 30 years. His general physical examination revealed single non tender 1.5×1 cm lymphadenopathy left station IB with normal

overlying skin. Local examination revealed 5×5 cm black-coloured mass with irregular margins on left side crossing midline starting from gingivobuccal margin to left sided hard palate. Systemic examination was with in normal limits.

CECT BOS to T4 revealed heterogeneously enhancing soft tissue mass 5×4.3×3.4 cm involving left maxillary alveolar ridge and adjacent gingivobuccal space and hard palate on left side. IHC from biopsy blocks were positive for S100, SOX10, HMB45 and negative for CK with Ki67 60-70% (Figure 2 a-d).

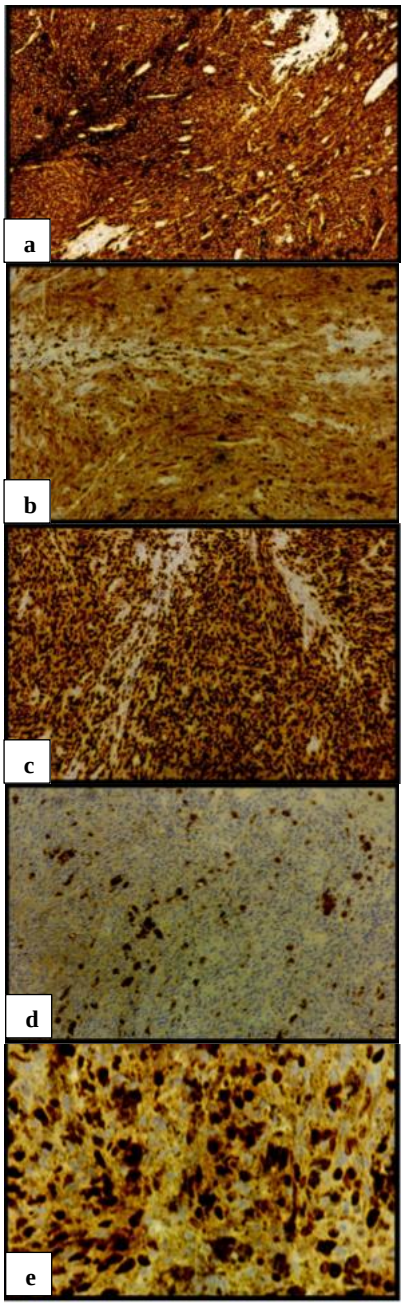

Figure 2: (a) IHC showing positivity for HMB45; (b) IHC showing positivity for S100; (c) IHC showing positivity for SOX10; (d) IHC negative for CK; (e) IHC showing high ki67 (MIB-1) 60-70%.

Patient was investigated and put on temozolamide and thalidomide resulting in drastic reduction in the size of the tumor after two cycles (Figure 3). He was planned for split course hypo-fractionated radiotherapy in view of his old age but he refused the further treatment and is on oral chemotherapy only which he is tolerating well without disease progression.

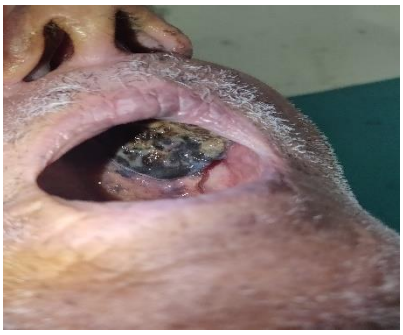

Figure 3: Case 2 showing drastic reduction post two cycles of oral TMZ and thalidomide.

DISCUSSION

Malignant melanoma is an aggressive malignancy with extremely high chances of metastasis occurring as a result of excessive proliferation of malignant melanocytes. Oral cavity melanomas are thought to arise de novo in contrast to cutaneous counterparts which are etiologically linked to exposure to the sun. Incidence is less accounting for only 0.5% of oral malignancies and most frequent sites involved are maxillary gingiva and hard palate . Mucosal melanoma of the head and neck have been known to carry worse prognosis.[5] The first patient had unusually aggressive course of the disease partly due to break in adjuvant treatment after resection and partly due to COVID-19 pandemic restrictions because of which he kept on shifting from one centre to another. Aftermath was a gigantic growth invading almost entire mouth opening resistant to almost every line of chemotherapy and radiation therapy.

TMZ is very well tolerated and improves the quality of life of patients with metastatic melanoma.[6] Second patient presented with oral cavity MM in advanced age (82 years) reported a drastic reduction in size of the lesion and had subjective improvement after initiation of therapy. As patient refused radiotherapy treatment, he continued oral chemotherapy. He is having good quality of life with stable disease on last contact.

Histologically melanoma has a large spectrum of features which may mimic epithelial, hematologic, mesenchymal and neural tumors. Immunohistochemistry (IHC) has been the primary tool to differentiate melanomas from these other tumors. S-100 remains the most sensitive marker for melanocytic lesions. The other markers with good specificity are HMB-45, MART-1/Melan-A,

tyrosinase and MITF.[7] Both patients had IHC positive for S100 and HMB45.

Radiological imaging with CT scan mucosal melanomas may present as polypoid mass along with bony remodelling or erosion with strongly contrast enhancement. MRI may reveal homogenous signal intensity on T1 weighted image. High T1 signal may be seen secondary to haemorrhage or melanin with T2 low signal. Metastases return the same signal characteristics as the primary lesion.[8]

Differential diagnosis for this ominous neoplasm include benign melanocytic lesions, dysplastic nevus, blue nevus and pigmented spindle cell tumor.

CONCLUSION

Mucosal melanomas generally carry a worse prognosis compared to cutaneous melanomas. This makes early diagnosis (by means of thorough oral cavity examination with biopsy of suspicious lesion) and treatment important step to provide better prognosis for the patient. Multidisciplinary approach was mandatory to treat the tumors at early and locally advanced stages. Goal at advanced stages was to achieve a better quality of life and palliation of symptoms like pain and bleeding. General public awareness about oral self-examination and periodic oral check-up can aid to detect such notorious neoplasms in their infancy.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

REFERENCES

1. Hicks MJ, Flaitz CM. Oral mucosal melanoma: epidemiology and pathobiology. Oral Oncol. 2000;36(2):152-69.
2. Kumar V, Vishnoi JR, Kori CG, Gupta S, Misra S, Akhtar N. Primary malignant melanoma of oral cavity: a tertiary care center experience. Natl J Maxillofac Surg. 2015;6(2):167-71.
3. McLaughlin CC, Wu XC, Jemal A, Martin HJ, Roche LM, Chen VW. Incidence of noncutaneous melanomas in the U.S. Cancer. 2005;103(5):1000-7.
4. Ebenezer J. Malignant melanoma of the oral cavity. Indian J Dent Res. 2006;17(2):94-6.
5. Hahn HM, Lee KG, Choi W, Cheong SH, Myung KB, Hahn H. J. An updated review of mucosal melanoma: survival meta-analysis. Mol Clinic Oncol. 2019;11(2):116-26.
6. Li RH, Hou XY, Yang CS, Lou W, Tang J, Liu Y, et al. Temozolomide for treating malignant melanoma. J Coll Physicians Surg Pak. 2015;25(9):680-8.
7. Ohsie SJ, Sarantopoulos GP, Cochran AJ, Binder SW. Immunohistochemical characteristics of melanoma. J Cutan Pathol. 2008;35(5):433-44.
8. Radiopaedia. Fact sheet: Sinonasal mucosal melanoma Available at: https://radiopaedia.org/articles/sinonasal-mucosal-melanoma. Accessed 17 December 2020.

Cite this article as: Sharma J, Gupta M, Saini A. Malignant melanoma oral cavity: case report of two patients from sub- Himalayan region. Int J Otorhinolaryngol Head Neck Surg 2021;7:896-9.