

Unusual Metastasis and Presentation in a Case of Carcinoma Lower Alveolus: A Case Report

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Abstract

Bony metastasis from head and neck carcinoma are exceedingly rare. In India, head and neck carcinoma often presents at a locally advanced stage but still bony metastasis are rare. Here, we mention a case which presented with symptoms of bone pain and destruction and on examination we found a primary lesion in the buccal mucosa.

Our case report reflects a rare presentation with a unusual site of metastasis along with the management of such rare presentation of the disease.

Keywords: Unusual presentation, Bone metastasis, Head & Neck Cancer.

INTRODUCTION

Carcinoma of head and neck region is the most common cancers worldwide but is more common in Asian countries. It is 2% of all cancers incidences around the world. As per Globocan 2022 data, worldwide incidence of oral cancers was 389,485 and new cases comprising of 1.9% of all cancers among which 60% present in locally advanced stage. As per the Indian Council of Medical Research, 0.2-0.25 million new head and neck cancers are diagnosed every year. In 2022, Globocan reported 143,759 new cases of oral cavity cancers in India with two-thirds male and 79,979 deaths.¹ Histologically, 90% of head and neck cancers are squamous cell carcinomas, while remaining are adenocarcinoma, lymphomas, sarcomas and very few of some rare pathology. The usual spread is

either local spread or to the lymph nodes. Distant metastasis is usually rare. Oral Tongue and buccal mucosa are more common sites involved in the Indian subcontinent.

High incidence of head and neck cancers (HNC) in India is mainly attributable to consumption of oral tobacco, betel leaves, and nuts along with lime. Most of the cases present with advanced stage probably due to lack of awareness, poor socio-economic condition or non-availability of diagnostic facilities in rural areas. The advanced stage at presentation is responsible for loco regional failure and distant metastasis. Few studies have reported distant metastasis rates between 4% and 26% in patients with most common histology of squamous cell carcinoma. Most common sites for distant metastasis is lung followed by liver, mediastinum, and rarely can involve bone and brain. Bone metastasis (BM) is comparatively infrequent in

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head and neck squamous cell carcinoma (HNSCC) and has survival of around 6-months only, after development of metastasis. BM also decreases the quality of life in such patients.

CASE REPORT

A 66 year old male, hypertensive, non-diabetic presented with a complaint of a soft tissue swelling, which was non tender, presented over the occipital region of scalp and associated with back pain. Further on examination of oral cavity, an ulcer was present over the left buccal mucosa. (Fig. 1) Histopathological examination of the ulcer was suggestive of well differentiated Squamous cell carcinoma. Whole Body PET CT Scan (Fig. 2, 3) was suggestive of 3.9 x 1.8 cm lytic lesion involving left mandibular alveolus involving molar / retromolar region extending upto mandibular ramus involving lower gingiva buccal sulcus (GBS). Multiple FDG avid (SUV max 4.6 - 7.3) sub cm sized bilateral cervical level II, level III, left cervical level IV, level V lymph nodes were also seen. There was a lesion present involving superolateral wall of right orbit which is abutting the right eyeball and another scalp lesion of size 5x5 cm was seen. In order to rule out metastasis in brain, an MRI BRAIN was done which showed multiple well defined variable sized calvarial marrow lesions of which largest was located at the roof of right orbit and another scalp lesion along the left occipital bone with bony erosion. FNAC samples were collected from

the scalp swelling and the swelling present over the right supra orbital region (which developed in the course of presentation). The cytological examination was suggestive of Metastatic Carcinoma. Haematological examination showed high levels of creatinine and severe anemia, hence conservative treatment was advised. After recovery, the patient was to be planned for palliative aim chemotherapy in view of metastatic disease.



Fig. 1: A. Lesion left lower alveolus, B. Swelling Left parotid Area, C. Swelling Left Occipital area

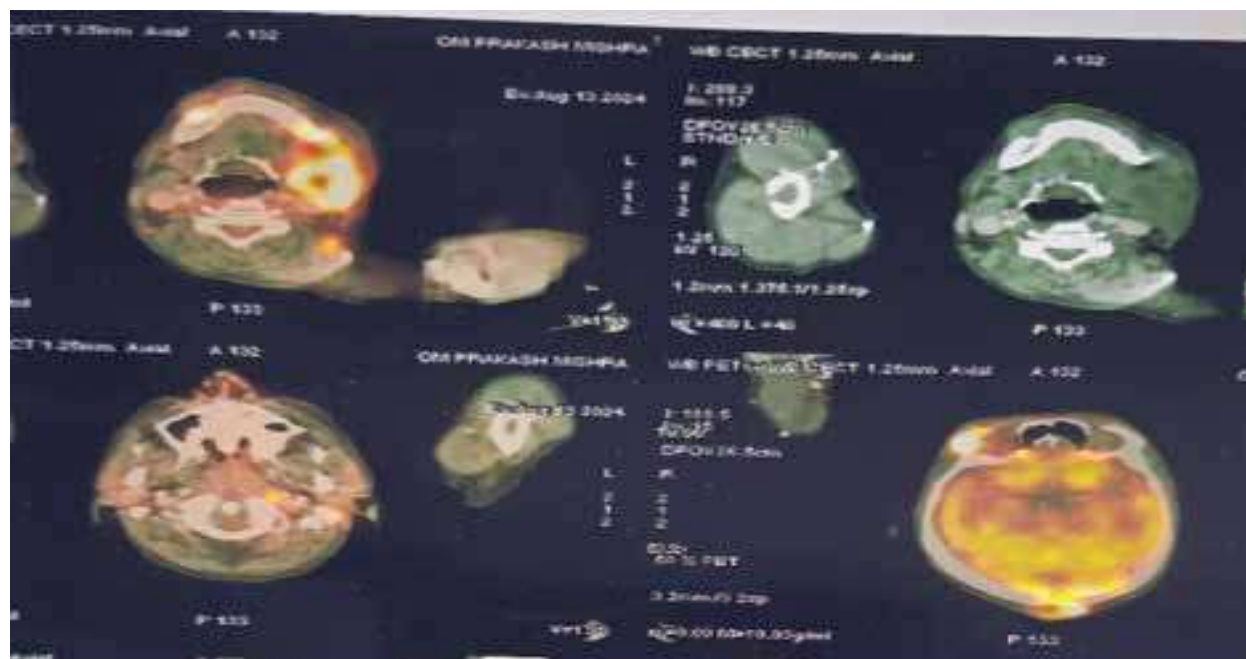


Fig. 2: PET showing high uptake in Parotid and Occipital area

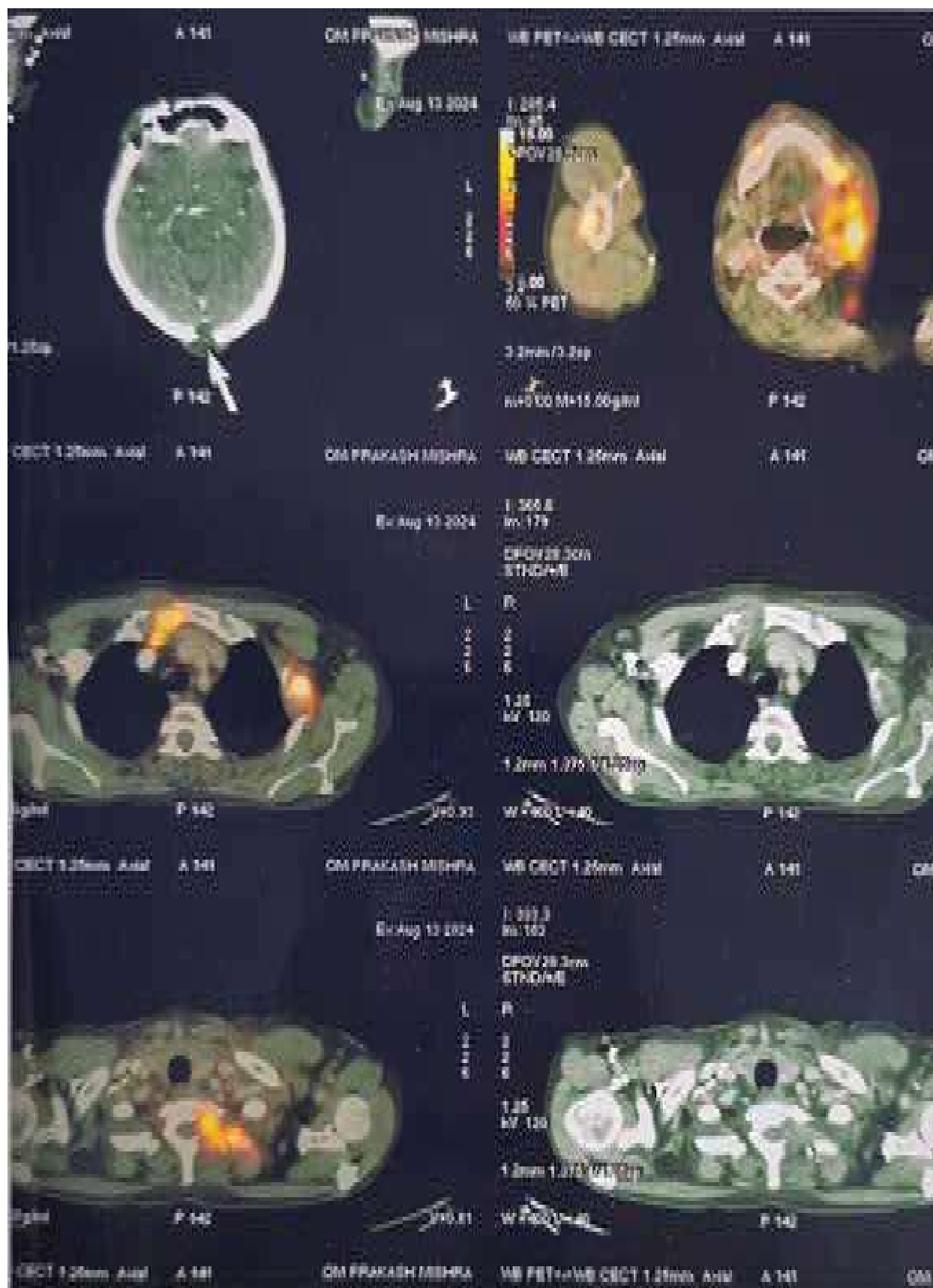


Fig. 3: PET showing high uptake in Left Lower alveolus, Vertibrae, axilla and sternal area

DISCUSSION

Lungs is the most common site of distant metastasis in HNC, followed by cervical lymph nodes, bone, liver, and skin.² Bone metastases has generally been considered a rare and late event in patients with head and neck cancer with reported incidence varying with tumor site involvement, ranging from 50%-80% in patients with nasopharyngeal carcinoma (NPC), while only 2%-22% in squamous cell carcinomas of other head and neck sites.^{3,4} The prognosis of HNC patients with bone metastases is reportedly poor with an expected median overall survival of about 8 months.⁵ Bone involvement greatly diminishes quality of life since it causes severe bone pain and skeletal related events (SREs) such as bone fractures, spinal cord compression, and hypercalcemia. Head and neck carcinoma generally spreads via lymphatics and only 15-20% have non-lymphatic spread. The frequency of osseous dissemination in HNC depends greatly upon the primary tumor size (T) and the regional nodal (N) involvement. T and N staging of patients at presentation also affects the prognosis.⁶

It is reported that the primary site (hypopharynx vs. other sites), size more than 2 cm (T2 & above), tumor grading and nodal status (more in N3 node), disease with extra-capsular extension (high prevalence) and prognostic stage (higher incidence in stage IV disease than others) are the major contributory risk factors for the development of distant bone metastasis.⁷ As far as the primary site of the tumor is concerned, the prevalence of distant metastasis in bone as well as other organs, is mostly seen in the tumor of the hypopharynx, followed by oropharynx (base of tongue)⁸ Bhandari *et al.* reported that among different primary sites of head and neck tumors, the hypopharynx is more likely to develop distant metastases with a probability of 20.5-60% and has a poor prognosis.⁹

Bone metastases are recognized with symptoms and then by imaging modalities such as bone scintigraphy, plain X-rays, magnetic resonance imaging/computed tomography (CT), and positron emission tomography/CT (PET/CT) and confirmation is done by biopsy or fine needle aspiration cytology. León *et al.* have shown that advanced local and nodal disease with lymphovascular invasion, extracapsular spread in three or more lymph nodes have increased chances for the development of distant metastases especially when jugular vein is involved.¹⁰ Eighty

five percent of patients of breast, prostate, or lung cancer are associated with bone metastases. Some lesser extent of primary sites with a predisposition for bone metastases include thyroid, melanoma and kidney.

Bone metastases develop most commonly in axial skeleton including spine, pelvis and ribs. Lumbar spine is the most common site to get involved as seen in the previous literature.⁹ The spread of head and neck cancers to bone is probably because of the tumor cells systemically spreading along the sites of red bone marrow distribution in the skeletal system.¹¹ Most often metastasis is through the hematogenous route or by direct extension. The bone metastasis can be osteolytic or osteosclerotic or mixed but most commonly in head and neck cancers it is osteolytic. The mechanism of pain from bone metastases can be mechanical instability of the bone, irritation of periosteal stretch receptors, tumor directed osteoclast mediated osteolysis, tumor-induced nerve injury, production of nerve growth factor or stimulation of cytokine receptors or the tumor cells themselves. The study of Bhandari and Jain showed that 6 (0.96%) out of 624 patients of head and neck cancers cases developed metastasis to single or multiple bones during the disease, with buccal mucosa and tongue primary seen in most of the cases.⁹ Distant metastasis has a greater impact on survival, so early detection can regulate therapeutic strategy. For high-risk patients, routine metastatic work-up for lung, liver and bone should be performed and any type of mild bone pain should not be left un-investigated. León *et al.* demonstrated 37%-57% prevalence of bone metastases at autopsy which is much higher than in clinical studies i.e., 4%-26%.¹⁰

A retrospective study by Pietropaoli *et al.* was conducted using radiographs and nuclear medicine studies on 363 cases of head and neck squamous cell carcinoma to identify the bone involvement and showed that 1% of these patients had bone metastases. Bone involvement was identified in eight sites, three in pelvis, two in femur and one each in humerus, rib and thoracic spine. Lesions were mostly lytic and very few were blastic. Time from initial primary tumor diagnosis to the recognition of metastatic lesions ranged from being present at diagnosis to a maximum of 3.5 years. Time from the identification of metastatic lesions to patient death was not more than 8 months. All bone lesions had appeared 3-12 months after the completion of primary treatment.⁷ In other studies, the observation remained the same regarding the site and duration. In the study by Bhandari *et al.* the

bone metastasis occurred 9 months to 3 years after completing the primary treatment and patients survived from 3 to 11 months with solitary or multiple bone metastases.⁹

Bony involvement by distant metastasis in HNC is crucial in clinical practice because they cause a lot of misery to patients, including severe refractory pain, pathological fractures, spinal cord compression and hypercalcemia & associated complications. Palliation with the help of radiation and salvage chemotherapy are the routine treatment planned for the patients with HNSCC with distant organ involvement. Platinum-based systemic chemotherapy has shown some benefit and improves the outcome to a certain degree.¹² Radiotherapy to the involved bone, either single session or multi-fractionation regimen is usually employed in all BM patients, along with systemic chemotherapy or bone directed treatment therapy by bisphosphonates (viz. zoledronic acid).¹² Once BM develops in these patients, median survival time is significantly decreased.

Hence, the information regarding the natural history of various tumors & the factors that are associated with distant metastases as well as good clinical judgment are essential for the cost-effective approach of treatment planning and decision making for both pre and post-operative assessment of distant metastases in head and neck cancer.

CONCLUSION

The unusual presentation of bone metastasis has led to the consideration for degree of dissemination of the primary disease in case of a Head and Neck carcinoma. The treatment plan is systemic chemotherapy, as well as local radiation for pain control in such metastatic disease. With newer techniques and treatment modalities, the increase in the overall survival in patients is leading to increased incidence of bone metastases and during follow up it should be considered a possibility in any patient with a concurrent or past diagnosis of head and neck carcinoma. The survival from the identification of bone metastasis to death in these patients is very low. Hence, patient should be investigated during follow-up considering possibility of distant osseous metastases.

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