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Mandibular Osteosarcoma: A Rare Case Study Highlighting the Importance af AaMultidisciplinary Approach

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ABSTRACT:
Osteosarcoma is a malignant mesenchymal tumour characterized by the production of osteoid matrix by neoplastic cells. While commonly affecting the long bones, osteosarcoma of the jawbones is a rare entity, accounting for less than 10% of all osteosarcomas. Here we present a case report of 50 a year old female, who presented to our institute with a chief complaint of pain and swelling in the right lower one-third of their face for 2 weeks. Emphasizing the clinical, radiological, and histopathological features, we diagnosed the lesion as osteosarcoma. Osteosarcoma of the oral cavity is a rare and aggressive malignancy that often presents with nonspecific symptoms, leading to delayed diagnosis. Radiographic features such as mixed radiolucent-radiopaque lesions and a sunburst appearance can aid in early detection. Histologically, characterized by malignant mesenchymal cells producing osteoid. The objective of presenting this case is that osteosarcoma is a rare but aggressive tumour that requires a high index of suspicion and timely diagnosis.

Keywords: Osteosarcoma, oral cavity, mandible, jaw tumour, sunburst, osteoid matrix

Introduction

Osteosarcoma is a rare, aggressive malignant tumour of mesenchymal origin, defined by the presence of malignant cells producing osteoid or immature bone. While it is the most common primary malignancy of bone, its occurrence in the craniofacial skeleton, particularly within the jawbones, is an infrequent phenomenon, accounting for approximately 6–8% of all osteosarcomas and less than 1% of head and neck malignancies.[1] Osteosarcomas of the jaws demonstrate several clinicopathological features that are distinct from those of their appendicular counterparts.

The mandible is more frequently affected than the maxilla, often involving the posterior body and ramus region. There is a slight male predilection, typically in the third to fifth decades of life. [2]

Management of jaw osteosarcoma is challenging and mandates a multidisciplinary approach. Despite the relatively lower rate of distant metastasis compared to long bone osteosarcomas, local recurrence is a significant concern in the maxillofacial region due to complex anatomy and difficulties in achieving wide surgical clearance.[3] The overall prognosis of gnathic osteosarcomas is better than that of

conventional OS, with reported 5-year survival rates ranging from 40% to 70%, depending on tumour size, location, histologic grade, and adequacy of surgical margins.

CASE PRESENTATION

This case report aims to highlight the clinical, radiographic, and histopathological features of osteosarcoma in the oral cavity, emphasizing the importance of early diagnosis and appropriate management. We present a rare instance of mandibular osteosarcoma in a 50-year-old female.

A 50-year-old female patient presented to our institute with a chief complaint of pain and swelling in the right lower one-third of their face since 2 weeks. The patient gives a history of pain, which is moderate and gradual in onset, intermittent, and radiating in nature.

Extraoral examination showed a solitary diffuse swelling on the right side of the face involving the lower one-third of the face. The swelling was 4x4.5cm in size and ovoid in shape, anteroposteriorly extending 2cm away from the lateral side of the angle of the mandible to 1cm lateral to the mental region, superior-inferiorly 1cm short of the ala of the nose extending up to the inferior border of the mandible. (fig.1)

Intraorally, a solitary, well-defined, firm to hard swelling was noted in the right posterior mandibular alveolar region, extending from the 44 to 47. (fig.2) The swelling measured approximately 3 × 3 cm and exhibited buccolingual cortical expansion, more prominent on the buccal aspect, resulting in slight obliteration of the buccal vestibule. On palpation, the swelling was non-tender, non-compressible, and non-fluctuant. The adjacent gingiva appeared slightly erythematous.

Fig .1. Clinical picture showing extraoral swelling at angle of mandible

Fig. 2. Clinical picture showing mass extending from 44 to 47

CBCT imaging of the mandible was performed to assess the extent of the lesion and its effect on surrounding structures. The scan revealed a poorly defined, mixed radiolucent-radiopaque lesion located in the right posterior mandibular body. The "classic" sunburst or sunray appearance caused by osteophytic bone production on the surface of the lesion is appreciated on an occlusal projection.

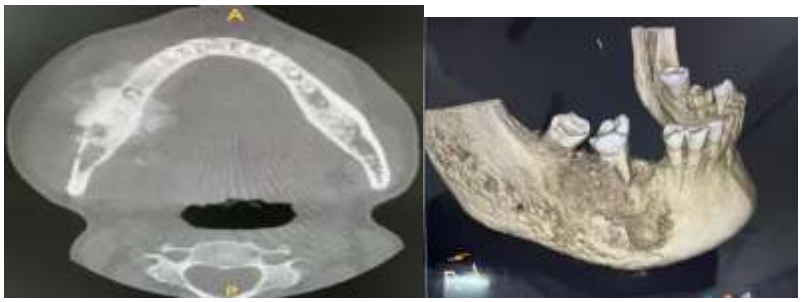

Fig. 3. CBCT showing classic sunburst appearance

Based on clinical examination and radiographic findings, a provisional diagnosis of Osteosarcoma was considered.

Excisional biopsy was carried out under local anaesthesia and the specimen was obtained, specimen was firm to hard in consistency with bony spicules.

Fig. 4. Gross specimen received showing massive tumour growth

Microscopic examination revealed the presence of neoplastic cells arranged in sheets, nests, and trabecular patterns within the tumour stroma. A prominent production of thin, lacy osteoid material in the stroma was noticed. Foci of lamellar bony trabeculae were seen within the tumour mass. The tumour cells were pleomorphic, with marked variability in cellular and nuclear size and shape. Hyperchromatic nucleus with prominent nucleoli, and the cytoplasm was eosinophilic. Mitotic figures were noticed at focal areas. The osteoid was irregular and haphazardly deposited. Focal rimming of osteoid by osteoblast-like cells was evident.

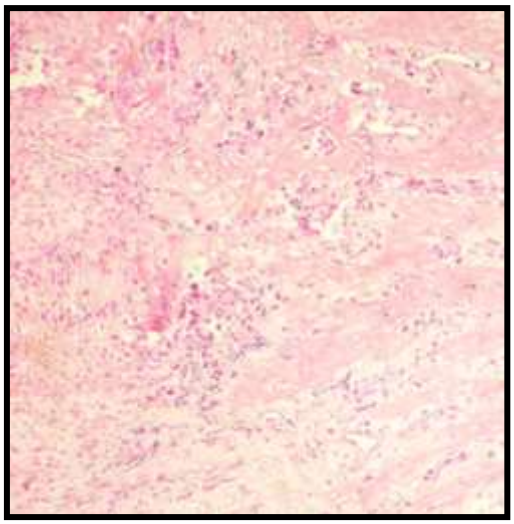

Fig. 5. Photomicrograph of Osteosarcoma (4x)

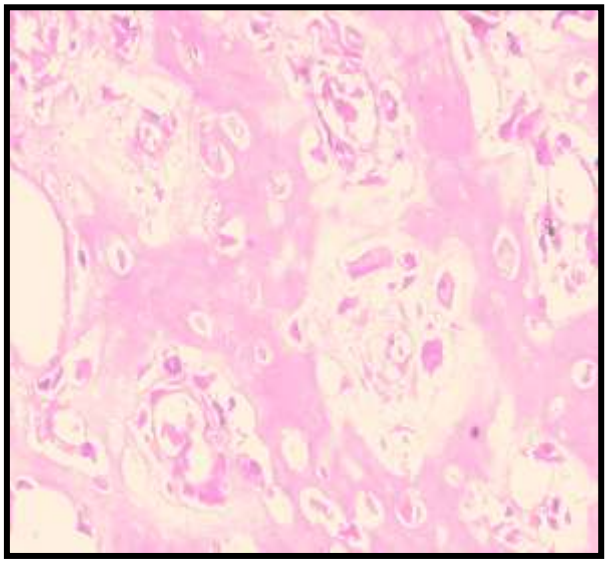

Fig.6. Photomicrograph showing dysplastic ossifications and lacunae filled with osteoblasts showing hyperchromatic nuclei (40x)

Based on clinical, radiographic, and histopathological features, a final diagnosis of Osteosarcoma was made.

The patient underwent segmental mandibulectomy with clear surgical margins and was sent to higher centres for further treatment procedures.

DISCUSSION:

Osteosarcoma (OS) is a rare but aggressive malignant bone tumour characterized by the direct formation of osteoid by malignant mesenchymal cells.[1] While it most frequently affects the metaphyseal regions of long bones in adolescents and young adults, its manifestation in the jawbones is an uncommon clinical entity, accounting for less than 10% of all osteosarcomas and <1% of head and neck malignancies.

The pathogenesis of osteosarcoma involves complex genetic and molecular alterations that lead to the malignant transformation of mesenchymal stem cells into osteoblast-like cells capable of producing immature bone matrix (osteoid). Several key molecular pathways and genetic mutations have been

implicated. RB1 (Retinoblastoma gene) and TP53 (tumor protein p53) mutations are among the most commonly identified alterations in OS.[4] Loss of tumor suppressor activity in these genes promotes unchecked cellular proliferation and genomic instability. MDM2 amplification, CDK4 overexpression, and abnormalities in the PI3K/AKT/mTOR and Wnt/β-catenin signalling pathways have also been associated with osteosarcoma pathogenesis. In jaw osteosarcomas specifically, the exact aetiology remains unclear. However, predisposing factors such as prior radiation therapy, Paget's disease, fibrous dysplasia, and trauma have been proposed, although most cases remain idiopathic. Unlike conventional OS, jaw osteosarcomas are typically of lower histological grade, and have less frequent metastasis, but are prone to local recurrence due to challenges in achieving adequate surgical margins in the complex anatomy of the maxillofacial region.[5]

Clinically, jaw OS may manifest with facial swelling, pain, tooth mobility, malocclusion, paraesthesia, or, less commonly, mucosal ulceration.[6] Intraorally, the lesion in our case presented as a firm, non-ulcerated swelling with cortical expansion, mobility, and buccal vestibular obliteration—findings that may be easily mistaken for more benign odontogenic lesions such as ossifying fibroma, cemento-ossifying dysplasia, or chronic osteomyelitis.[7]

Radiographically, jaw osteosarcomas may appear as radiolucent, radiopaque, or mixed-density lesions with ill-defined, invasive margins.[8] The presence of a "sunburst" periosteal reaction is a classic feature, often resulting from rapid, disorganized periosteal bone formation.[9] In our case, CBCT revealed a poorly defined, mixed radiolucent-radiopaque lesion located in the right posterior mandibular body. The "classic" sunburst or sunray appearance caused by osteophytic bone production on the surface of the lesion is appreciated on an occlusal projection.

Histologically, osteosarcoma of the jaw is classified into several subtypes based on the predominant matrix: osteoblastic, chondroblastic, and fibroblastic.[10] The hallmark diagnostic criterion remains the presence of malignant osteoid directly laid down by anaplastic stromal cells.[11] The osteoblastic variant, as seen in our case, is the most frequently encountered subtype and is characterized by a dense, irregular osteoid matrix interspersed with pleomorphic malignant osteoblasts.[12] However, diagnosis can be challenging and often requires correlation with clinical and radiographic data due to overlap with benign fibro-osseous lesions and low-grade malignancies.

The differential diagnosis for jaw osteosarcoma includes: Fibrous dysplasia, Ossifying fibroma, Paget's disease, Chronic sclerosing osteomyelitis, Cemento-osseous dysplasia, Ewing's sarcoma, and Chondrosarcoma.[11, 13]

Differential Diagnosis	
Aneurysmal bone cyst • Benign, but can be large and expansile • May be secondary to other lesions (eg, chondroblastoma, fibrous dysplasia, giant cell tumor) • No cytologic atypia, lesional necrosis or atypical mitotic figures • Admixed inflammatory cells • Osteoid often present parallel to cyst wall and not associated with atypical cells	Fibrous dysplasia • Fibrous stroma with plump, oval to elongated fibroblasts with smooth nuclear outlines and no significant cytologic atypia • Usually thinner, shorter or more delicate trabeculae of woven bone • No permeation of surrounding tissues Desmoplastic Fibroma • Similar to soft tissue desmoid fibromatosis • No bone or osteoid production by tumor cells
Ewing sarcoma/primitive neuroectodermal tumor • Usually smaller, more uniform cells than small cell OGS • No truly spindled tumor cells or cellular anaplasia • No osteoid or chondroid matrix production by tumor cells • Membranous CD99 staining • Presence of t(11;22) or *EWSR1* gene rearrangement	Giant cell tumor of bone • Epiphyseal location; skeletally mature patient • Reactive/metaplastic bone formation, numerous mitotic figures, lesional necrosis and lymphovascular invasion may be seen • Evenly placed osteoclast-like giant cells amid uniform mononuclear cells • No osteoid or chondroid matrix production by tumor cells, no anaplasia

The mainstay of treatment for mandibular osteosarcoma is wide surgical excision with tumour-free margins.[12] Due to the complex anatomic constraints of the maxillofacial region, achieving adequate oncologic clearance can be challenging. Adjuvant chemotherapy is often considered for high-grade tumours, although the evidence supporting its use is less robust compared to conventional osteosarcoma.[10] The role of radiotherapy remains limited due to the relative radioresistance of the tumour and the radiosensitivity of surrounding vital structures, although it may be employed in cases with close/involved margins or unresectable disease.[5]

Prognostically, jaw osteosarcomas tend to have a better overall survival than their long-bone counterparts, with 5-year survival rates ranging between 40% and 70%.[1] Factors influencing prognosis include tumour size, location, histologic grade, surgical margins, and the presence of local recurrence. Metastasis typically occurs in the lungs.

CONCLUSION:

Osteosarcoma of the oral cavity is a rare but aggressive tumour that requires a high index of suspicion for timely diagnosis. Histopathological evaluation remains the gold standard for confirming a diagnosis, with recognition of malignant osteoid being crucial. Early detection, accurate pathological assessment, and a multidisciplinary treatment approach are vital for improving patient outcomes and minimizing the risk of recurrence.

References:

1. Chrcanovic BR, Gomez RS. Osteosarcoma of the jaws: A review of literature. *Oral Maxillofac Surg*. 2016;20(4):349-356.
2. Clark J, Unni KK, Dahlin DC, Devine KD. Osteosarcoma of the jaw. *Cancer*. 1983;51(12):2311-2316
3. Chaudhary M, Chaudhary SD. Osteosarcoma of jaws. J Oral Maxillofac Pathol. 2012 May;16(2):233-8. doi: 10.4103/0973-029X.99075. PMID: 22923896; PMCID: PMC3424940.

4. Slootweg PJ, Müller H. Osteosarcoma of the jaws: Analysis of 15 cases. *J Maxillofac Surg*. 1985;13(4):158-166.
5. Van Es RJ, Keus RB, Vermey A, et al. Osteosarcoma of the head and neck: The Netherlands Cancer Institute experience. *Head Neck*. 2002;24(9):894-900.
6. Nouri H, Mighri M, Ghorbel A, et al. Osteosarcoma of the mandible: A case report and review of literature. *Int J Surg Case Rep*. 2019;55:169-173.
7. Mardinger O, Givol N, Talmi YP, Taicher S. Osteosarcoma of the jaw: The Chaim Sheba Medical Center experience. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 2001;91(4):445-451.
8. Bianchi SD, Boccardi A, Di Alberti L, Saino M, Rizzatto G. Radiographic findings in osteosarcoma of the jaws. *Dentomaxillofac Radiol*. 2002;31(4):228-233
9. Kalia V, Kalra N, Bhalla AS, Sood R. Imaging of maxillofacial osteosarcoma: A pictorial essay. *Indian J Radiol Imaging*. 2012;22(3):195-200.
10. Eder-Czembirek C, Czembirek C, Selzer E, Lechner M, Thurnher D. Osteosarcoma of the mandible. *Oral Surg Oral Med Oral Pathol Oral Radiol*. 2013;115(6):e37-e43.
11. Neville BW, Damm DD, Allen CM, Chi AC. *Oral and Maxillofacial Pathology*. 4th ed. St. Louis: Elsevier; 2016.
12. Rajendran R, Sivapathasundharam B, editors. *Shafer's Textbook of Oral Pathology*. 8th ed. New Delhi: Elsevier; 2016.
13. Gonzalez AL, Cates JM. Osteosarcoma: Differential Diagnostic Considerations. Surg Pathol Clin. 2012 Mar;5(1):117-46. doi: 10.1016/j.path.2011.07.011. Epub 2011 Dec 15. PMID: 26837918.