

# Osteosarcoma of mandible: Detailed radiographic assessment of a case

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## Abstract

Osteosarcoma (OS) is a malignant connective tissue tumor originating from bone and is the most common primary bone malignancy of long bones but seldom arises in jaw bones. Osteosarcoma of jaws is frequently seen arising in the second and third decade as compared to earlier occurrences in other bones and show a slight predilection for body of mandible. It is a highly malignant tumor with varied radiographic features. We present a case with detailed radiographic assessment using intraoral radiograph, computed tomography (CT), 3-D CT, CT angiography techniques and histological evaluation.

**Keywords:** Computed tomography, osteosarcoma, osteosarcoma of jaws

## Introduction

Osteosarcoma (OS) is a mesenchymal malignancy of bone tissue. It accounts for 20% of all sarcomas and about 5% of all osteogenic sarcomas arise in the head and neck region.<sup>[1]</sup> It is the most common primary bone malignancy of skeleton other than craniofacial region. Primary OS in head and neck region is a rare occurrence and presents with distinct biologic behavior as compared to OS of long bones.<sup>[2]</sup> Osteosarcoma of jaws (OSJs) usually arise secondary to underlying pathologies such as fibrous dysplasia, Paget's disease, bone infarcts and therapeutic irradiation to craniofacial region.<sup>[3]</sup> Patients with OSJ usually report to dentists and is frequently misdiagnosed as periapical lesions, impactions or odontogenic lesions due to non-specific symptoms such as swelling, pain, tooth mobility, ill-fitting dentures and paresthesia.<sup>[4]</sup> Conventional radiographic features like Codman's triangle or widening of periodontal space are not pathognomonic of OSJ. Thus, radiographic techniques like computed tomography (CT) and magnetic resonance imaging (MRI) can be of great help in

assessing bone destruction and osteogenesis pattern. This may facilitate early diagnosis and treatment planning.

## Case Report

A 28-year-old male patient reported to Dental Department of a Private Hospital with the chief complaint of rapidly increasing swelling at left body of mandible [Figure 1]. Patient had visited an oral and maxillofacial clinic for tenderness at same site, and extraction of 38 was performed. After extraction, patient noticed fast progression of swelling with no relief in pain. Swelling was bony hard. Submandibular lymph nodes of involved side were palpable and non-tender. Intraoral examination revealed obliteration of buccal vestibule in relation to 35, 36 and 37. Intraoral radiograph, in early stage of disease, revealed widening of periodontal ligament (PDL) space in relation to 35, 36 and 37 without any evidence of odontogenic or periodontal disease.

Radiographic imaging using CT, 3-D CT and CT angiography techniques were performed. CT scan revealed a large tumor mass originating from surface of outer cortex of body and angle of mandible [Figure 2]. Lesion extended from first molar to posterior border of ramus antero-posteriorly. Tumor involved coronoid process above and extended 1.5 cm below lower border of mandible. Lower border of mandible was seen destroyed and replaced by connective tissue stroma of tumor. A mixed radiopaque and radiolucent mass with typical "sunburst" appearance at outer cortex with "codman's triangle" at the margins of tumor led to the diagnosis of OS [Figure 2a]. 3D CT images revealed extensive spread of tumor mass. Tumor osteoid was seen forming a complex network radiating from body and angle of mandible and reaching coronoid notch [Figure 3]. Study of bone destruction pattern on CT scans revealed a predominant bone-destruction negative pattern. At most areas of tumor involvement the continuity of outer cortical plate is intact, with tumor osteoid radiating outwards. However, at lower border of mandible tumor mass is seen destroying entire thickness of bone and replacing with radioluscent soft-tissue mass.

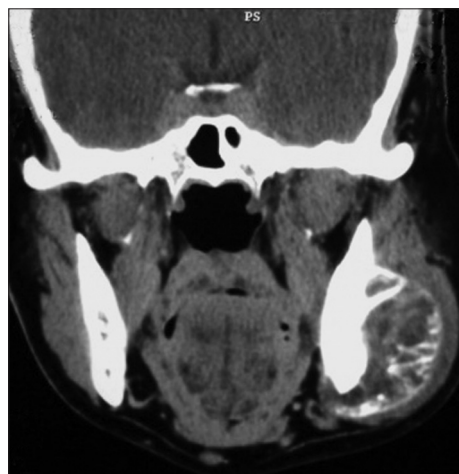
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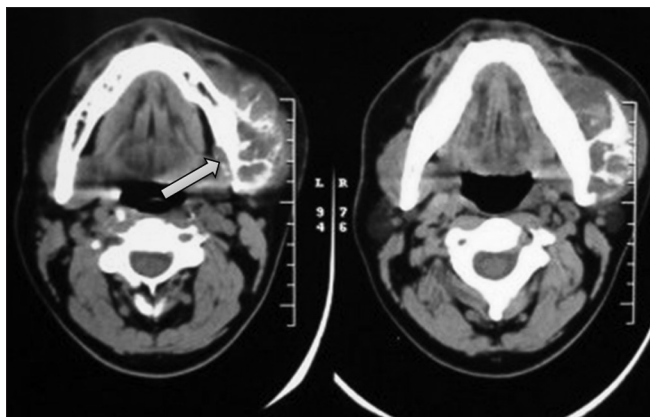
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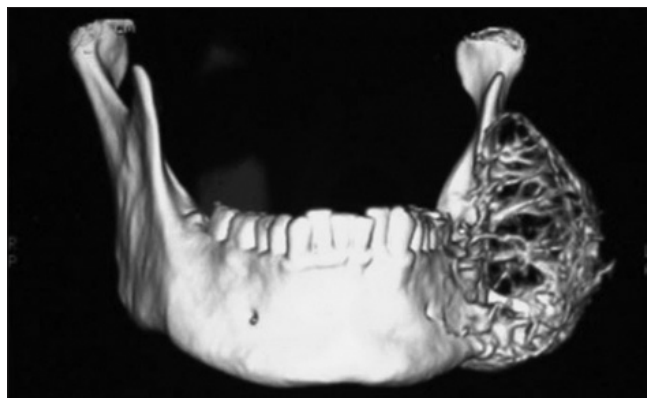
**Figure 1:** Clinical picture showing extraoral swelling at angle of mandible



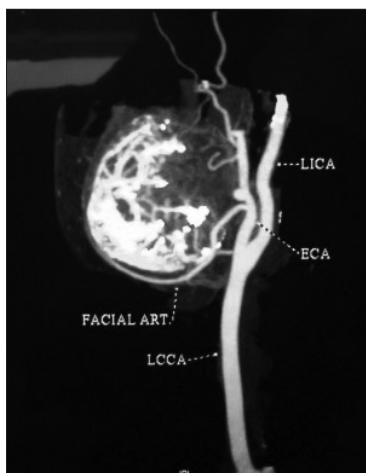
**Figure 2:** Axial computed tomography view showing bone spicules radiating perpendicular to outer cortex of mandible



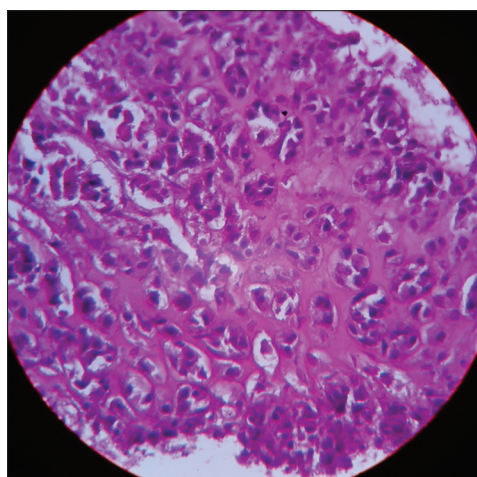
**Figure 2a:** Coronal computed tomography showing sunburst appearance of radiopaque mass from outer cortex, arrow shows area of cortical discontinuity



**Figure 3:** 3D-computed tomography image showing a dense meshwork of dysplastic bone



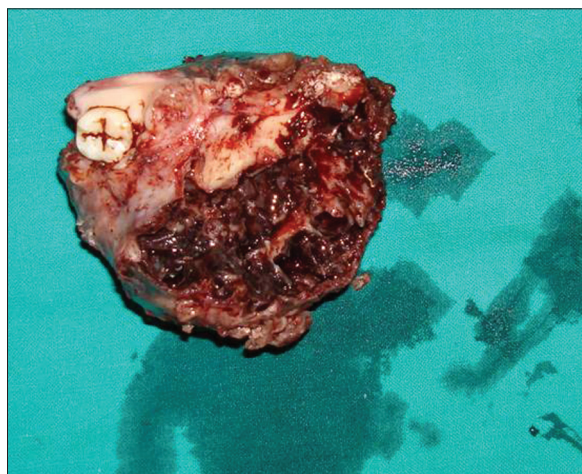
**Figure 4:** Computed tomography angiography showing arterial supply of tumor from external carotid artery



**Figure 5:** Photomicrograph showing dysplastic ossifications and lacunae filled with osteoblasts showing hyperchromatic nuclei

CT angiography was performed to assess the vascularity of tumor. The tumor was found to be extensively vascularized and the feeder vessel was identified to be external carotid

artery [Figure 4]. Incisional biopsy showed osteoid areas lined by plump osteoblasts with hyperchromatic nuclei, mitotic figures, and sheets of cells showing nuclear and cellular pleomorphism, inflammatory cells, numerous blood vessels and hemorrhage [Figure 5]. Case was operated upon under



**Figure 6:** Gross specimen received showing massive tumor growth involving angle and ramus of mandible

general anesthesia, tumor mass resection with reconstruction using iliac crest was done [Figures 6]. Final diagnosis was consistent with that of representative specimen.

## Discussion

Malignant tumors in jaws commonly presents with general symptoms such as pain, paraesthesia and swelling. Conventional radiographic techniques do not allow diagnosis and differentiation between benign fibro-osseous, inflammatory and malignant tumors. The radiographic differential diagnosis for OSJ includes fibrous dysplasia, osteomyelitis, osteoma, myositis ossificans and cement-osseous dysplasias. They all commonly arise in mandible, show mixed radiopacity and radiolucency and bony expansion with ill-defined border.<sup>[5-7]</sup>

OSJ is a highly destructive malignancy, which destructs cortical boundary. CT and MRI pictures may help in assessing the bone destruction pattern. MRI is more sensitive to identify both periosteal new bone deposition pattern and underlying bone abnormalities.<sup>[8]</sup> Clark *et al.*, classified radiographic pattern of OSJ into lytic, sclerotic and mixed. Osteolytic variants may show a completely radiolucent lesion. Sclerotic or osteolytic dominant pattern shows presence of radiopaque spicules in radiolucent mass. Osteogenic dominant pattern show areas of high density new bone formation.<sup>[9]</sup>

Panoramic radiograph is commonly available imaging modality and has a low radiation exposure. Though significant distortion and superimposition is seen, but a provisional differentiation of benign or malignant process can be made. It may clearly express bone remodeling, cortical destruction, tumor margins supero-inferiorly and antero-posteriorly.<sup>[10]</sup> CT is relevant in assessing tumor metastasis. CT in different planes can be used to create a 3D image, but radiation dose is much higher. MRI essentially helps to determine the extent of tumor invasion and involvement of any soft-tissue mass.

Not only it has high sensitivity for different radiodensities, it also avoids ionizing radiation.<sup>[5]</sup>

Symmetric widening of PdL space on panoramic radiograph is an important early feature of bone malignancies like in OS, chondrosarcoma and Ewing's sarcoma.

Study of bone destruction pattern and bone formation pattern can give an insight to the biologic behavior of disease. Destruction pattern of bone can be divided into two types. Bone destruction positive pattern shows discontinuity of cortex and bone destruction negative pattern may show expansion, but displays continuity of cortices.<sup>[5]</sup> Periosteal new bone formation is another important initial sign of bone disease. Osteogenesis pattern over CT can be a prognostic indicator. It can be divided into osteolytic and osteogenic patterns. Based on destruction and osteogenesis patterns CT findings can be classified in 4 headings, (1) osteolytic and bone destruction positive, (2) osteolytic and bone destruction negative, (3) osteogenic and bone destruction positive, and (4) osteogenic and bone destruction negative pattern.<sup>[11]</sup> Our case falls in 3<sup>rd</sup> category showing destruction of cortex with osteogenesis [Figure 2a].

It is very important to differentiate between different patterns of bone deposition seen in varying diseases. Periostitis show delicate periosteal elevation with curvilinear band of new bone. Layers of periosteal bone deposition in onion skin pattern are seen in Garre's osteomyelitis. Extensive new bone in layers parallel to cortex is seen in Ewing's sarcoma and marrow in filtrating diseases like leukemia. Solid homogenous new bone formation without lamellae is indicative of benign process such as langerhans cell histiocytosis, healing fractures, osteomyelitis and benign neoplasms. Spiculated linear opacities radiating out from cortex (sun burst) and Codman's triangle shows a discontinuous new bone formation indicating malignant process.<sup>[8]</sup>

Spicules of newly formed dysplastic osteoid may appear predominantly in a bone destruction negative pattern. It is indicative of fast biologic process. It represents a disturbance in reparative phase.<sup>[8]</sup> CT view shows a perpendicular alignment of dysplastic osteoid to jaw bones. This distinctive feature is called as "sun burst" appearance. This feature is seen in our case also.<sup>[5]</sup>

Periosteal reaction at periphery of tumor causing tent-like elevation of periosteum is seen in OS. New reactionary bone is soon destroyed by pathologic process leaving only a rim of new bone supporting periosteum. This tent-like raised periosteum with osteoid is termed as Codman's triangle. This is indicative of OS and sometimes also seen in fibrosarcoma and Ewing's sarcoma.<sup>[9]</sup>

Histopathological diagnosis of OSJ is based on pattern of unmineralized osteoid and mineralized bone tissue. The

intercellular osteoid matrix in OS appears eosinophilic, dense, homogenous material. The shape of osteoid is curvilinear with lacunae formation. Density and thickness of osteoid varies from flat and thick coalescing areas, to thinnest lace-like deposition between neoplastic cells referred as “filigree pattern.” Lacunae formed in dysplastic tumor mass shows presence of pleomorphic osteoblasts with hyperchromatic nuclei. At times lacunae appear empty and devoid of malignant cells due to “choking-off” and death of cells in central avascular portion of osteoid. Bone may appear eosinophilic or basophilic depending on amount of mineralization.<sup>[3]</sup>

Immunohistochemistry does not play a diagnostic role for OS. No specific marker to distinguish bone matrix from other collagenous counterparts is identified until date. Osteocalcin is the only protein exclusively produced by osteoblasts. It is helpful in distinguishing a primary bone malignancy from others. Alkaline phosphatase enzyme is strongly expressed in osteoblastic, chondroblastic and fibroblastic variants of OS but it can only be used over fresh cryostat sections or imprint sections. Negative staining for factor VIII and CD31 is an important feature expressed in OS. These immunohistochemical specificities can be helpful in diagnosing metastatic tumors and those with extensive anaplasia such as small cell or round cell variants.<sup>[12]</sup>

OSJs tend to spread locally within jaws. In mandible, nerves and foramen facilitate the spread of tumor to extragnathic sites. In maxilla cortices and foramen are thin and can be penetrated easily. Thus the disease in maxillofacial complex spreads to involve sinuses, nasal cavity, infratemporal fossa and other structures. Tumor invasion is usually found more than the clinical involvement. Spread through lymphatic route is rare and regional lymph nodes are relatively disease free in most of the cases.<sup>[2]</sup> Metastasis from mandible is far more common than maxilla.<sup>[3]</sup> Surgical exploration for biopsy, extraction of teeth and curettage procedures can cause extraosseous spread and metastasis. Treatment modalities include surgery, chemotherapy and radiotherapy. OSJs show best results with surgery with tumor-free margins. Radiotherapy significantly effects prognosis in skeletal OS, but holds no prognostic significance in OSJs. Chemotherapy has shown promising results both in recurrence incidences and survival, but does not improve poor prognosis of metastatic tumors. Chemotherapy is a prognostic indicator as it induces tumor necrosis and evaluation of tumor response can

be assessed.<sup>[3]</sup> Commonly used chemotherapeutic agents are cisplatin, doxorubicin, methotrexate, ifosfamide, etoposide.<sup>[13]</sup>

Misdiagnosis of OSJ by dentists, as tumors or cysts of odontogenic or non-odontogenic origin, leads to alternative treatments such as enucleation, marsupialization, curettage or segmental reactions. This only helps in spread of tumor and metastasis. Thus, knowledge of such highly malignant tumors is essential for early and adequate treatment.

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