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Case Report

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Atypical clinical presentation of mandibular osteosarcoma – a case report

Apresentação clínica atípica do osteossarcoma mandibular – um relato de caso

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ABSTRACT

Background: Osteosarcoma is a malignant bone tumor most commonly affecting the long bones, whereas involvement of the jaws accounts for approximately 6-9% of cases. The atypical clinical behaviour, diverse radiographic presentation and degree of abnormal cellular morphology often pose challenge to clinician in arriving at definitive diagnosis. Advanced imaging modalities like Computed Tomography (CT) and Magnetic Resonance Imaging (MRI) reveal better anatomical details than the conventional radiography. However, Fluoro deoxy Glucose Positron Emission Tomography/Computed Tomography (FDG-PETCT) offers greater accuracy, sensitivity, and specificity in diagnosing osteosarcoma as it provides the functional and metabolic information coupled with anatomical details **Objective:** To highlight the diagnostic value of FDG-PETCT as an adjunctive imaging modality in identifying an atypical presentation of mandibular osteosarcoma. **Case Report:** A 29-year-old female presented with a swelling in the right lower jaw for two months. Initial evaluation made by contrast enhanced CT and histopathological examination, following incisional biopsy were inconclusive. Subsequent assessment with FDG-PETCT coupled with immunohistochemical analysis of SATB2 facilitated in arriving at a definitive diagnosis. This enabled timely surgical intervention and appropriate management. **Conclusion:** FDG-PETCT plays a significant role in the early diagnosis, staging, and treatment planning of gnathic osteosarcoma. Therefore, it should be considered as an important adjunct in the diagnostic protocol for the management of osteosarcoma.

KEYWORDS

Bone neoplasms; Immunohistochemistry; Osteomyelitis; Osteosarcoma; Positron Emission Tomography Computed Tomography.

Resumo

Contexto: O osteossarcoma é uma neoplasia óssea maligna que afeta mais comumente os ossos longos, enquanto o envolvimento dos maxilares corresponde a aproximadamente 6-9% dos casos. O comportamento clínico atípico, a apresentação radiográfica diversa e o grau de morfologia celular anormal frequentemente representam um desafio para o clínico na obtenção de um diagnóstico definitivo. Modalidades avançadas de imagem, como a Tomografia Computadorizada (TC) e a Ressonância Magnética (RM), revelam melhor detalhamento anatômico do que a radiografia convencional. Contudo, a Tomografia por Emissão de Pósitrons com Fluorodesoxiglicose associada à Tomografia Computadorizada (FDG-PETCT) oferece maior acurácia, sensibilidade e especificidade no diagnóstico do osteossarcoma, ao fornecer informações funcionais e metabólicas associadas aos detalhes anatômicos. **Objetivo**: Destacar o valor diagnóstico da FDG-PETCT como modalidade de imagem adjunta na identificação de uma apresentação atípica de osteossarcoma mandibular. **Relato de caso**: Uma mulher de 29 anos apresentou aumento de volume na mandíbula direita, com dois meses de evolução. A avaliação inicial realizada por TC com contraste e exame histopatológico, após biópsia incisional, foi inconclusiva. A avaliação subsequente com FDG-PETCT, associada à análise imunohistoquímica

de SATB2, possibilitou a obtenção de um diagnóstico definitivo. Isso permitiu uma intervenção cirúrgica oportuna e um manejo adequado. **Conclusão**: A FDG-PETCT desempenha um papel importante no diagnóstico precoce e no planejamento do tratamento do osteossarcoma nos ossos gnáticos. Portanto, deve ser considerada um importante adjunto no protocolo diagnóstico para o manejo do osteossarcoma.

PALAVRAS-CHAVE

Neoplasias ósseas; Imuno-histoquímica; Osteomielite; Osteossarcoma; Tomografia por emissão de pósitrons combinada à tomografia computadorizada.

INTRODUCTION

Primary bone tumors account for about 0.2% of all cancers, which comprises of osteosarcoma (OS), Ewings sarcoma and chondrosarcoma [1]. OS is the most common primary malignant bony tumour affecting the long bones of adolescents and young adults [2]. OS of jaws is relatively rare and differs from its long bone counterpart, in terms age of onset, comparatively reduced propensity for metastasis and better overall survival rate [3]. Pain and swelling of the jaw are the most common symptoms and other associated signs include mobility of teeth, poor healing at the site of extraction, hypoesthesia or paraesthesia, especially in cases of tumor involving the mandible. Classical radiographic features include sunburst appearance, Garrington sign and Codman Triangle. Early diagnosis and early resection are the keys to better prognosis [4]. PETCT aids in the early diagnosis of OS by detecting heightened metabolic activity in bone lesions before structural changes are visible on conventional imaging. It assists in distinguishing benign from malignant lesions, identifying skip metastases, and offering whole-body assessment for early metastatic spread, thereby enhancing diagnostic accuracy and guiding treatment planning [5]. Treatment protocol also includes radical or conservative surgery followed by chemotherapy and radiotherapy. Here we report a case of a 29-year-old female whose histological appearance of the lesion closely resembled that of osteomyelitis but was eventually diagnosed as OS, and we review the role of PETCT in the early diagnosis of OS.

CASE REPORT

A female patient of late 20's reported to the Department of Oral Medicine and Radiology with a chief complaint of pain and swelling on her right lower jaw for the past 2 months. History revealed that patient underwent extraction two months back after which the swelling increased in size. The pain was dull, intermittent and aggravated on consuming food. Informed consent was obtained from the patient according to Declaration of World Medical association of Helsinki. Her medical and family history were non-significant with no history of any deleterious habits. Past dental history revealed patient underwent extraction of right mandibular third molar two months back after which patient noticed the swelling which was initially smaller in size and gradually increased to its current size. Extraoral examination revealed an ill-defined swelling in relation to the right angle of the mandible measuring approximately 2.0 × 1.5 cm in size with no evidence of discharge or secondary changes over the skin. On palpation, the swelling was hard to firm in consistency, mildly tender and fixed to the underlying bone with no evidence of abnormal cervical lymphadenopathy. Intraoral examination revealed mild erosions in relation to the alveolar mucosa of 47 and 48, along with poor wound healing at the extracted site of 47 and 48, as shown in Figures 1A and 1B. Radiographic evaluation using a mandibular lateral occlusal radiograph revealed a mixed radiopaque and radiolucent lesion on the right side of the mandible with lingual cortical expansion in relation to 47 and 48 regions, as shown in Figure 1C. The panoramic radiograph revealed ill-defined radiolucency along the right angle of the mandible in relation to 47 and 48 regions, denoting irregular areas of osteolysis as shown in Figure 1D. Periapical radiograph showed the presence of Garrington sign, characterized by symmetrical widening of the periodontal ligament space of the involved teeth. A contrast-enhanced CT (CECT) of the mandible revealed a large expansile lytic lesion measuring 4.4 × 2.8 × 4.5 cm over the angle of the right hemi mandible, involving the body and ramus of the mandible, along with a few areas of thinning and breaching of the cortical plate as shown in Figure 2. Based on the clinical and

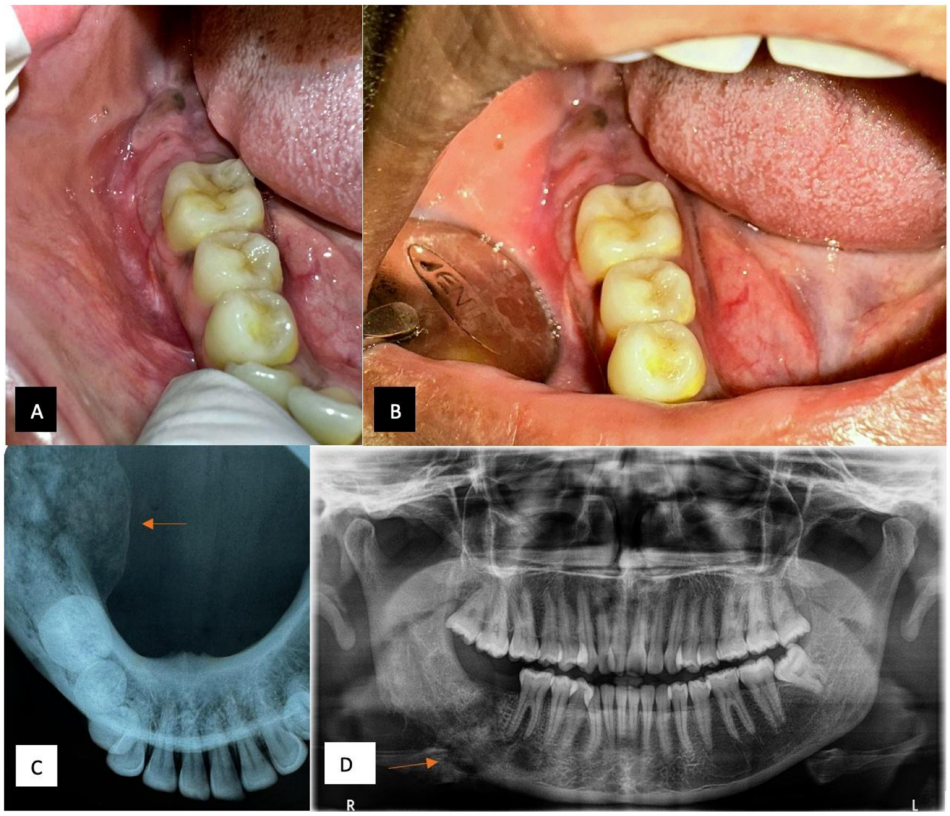

Figure 1 - (A) and (B) Intraoral picture shows erosions in relation to lower right angle of mandible; (C) Occlusal radiograph shows mixed radiopaque and radiolucent features with lingual cortical expansion in relation to 47 and 48 region; (D) OPG reveals an ill defined radiolucency in the right lower angle of mandible extending to the 48 and 47 region.

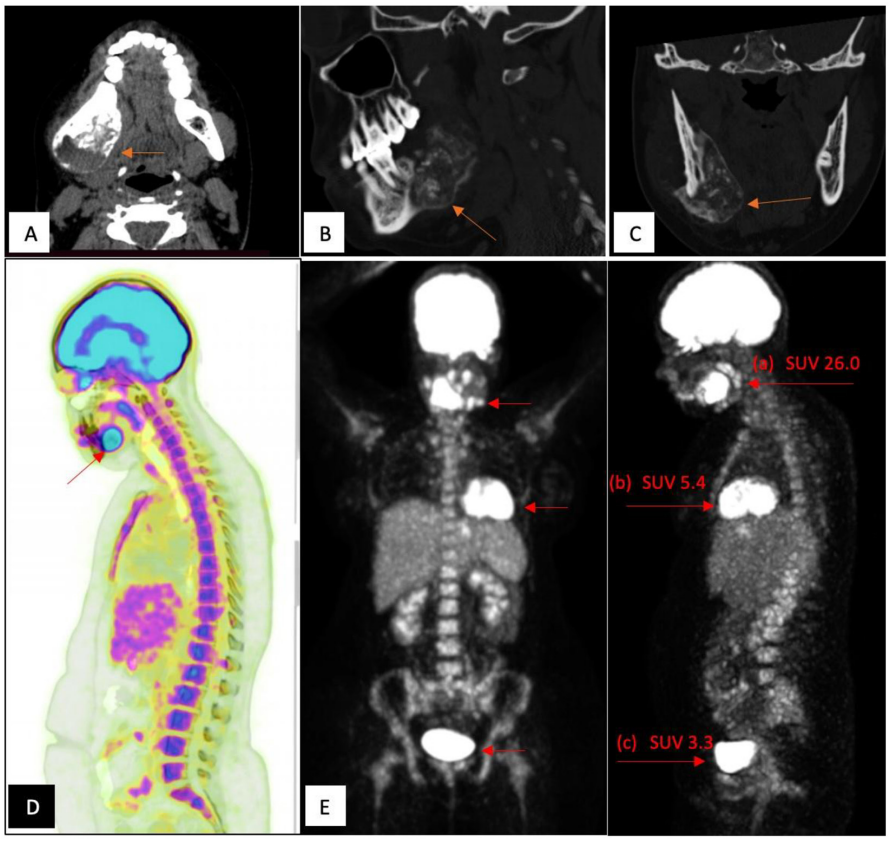

Figure 2 - (A-C) CT shows a destructive enhancing mass lesion involving the right angle of mandible; (D) and (E) - (a) PETCT shows a large expansile heterogenous lytic lesion centered over the angle of the right hemi mandible; (b) a hypodense lesion with centripetal enhancements of liver; (c) a diffuse perilesional node in the spine and pelvis.

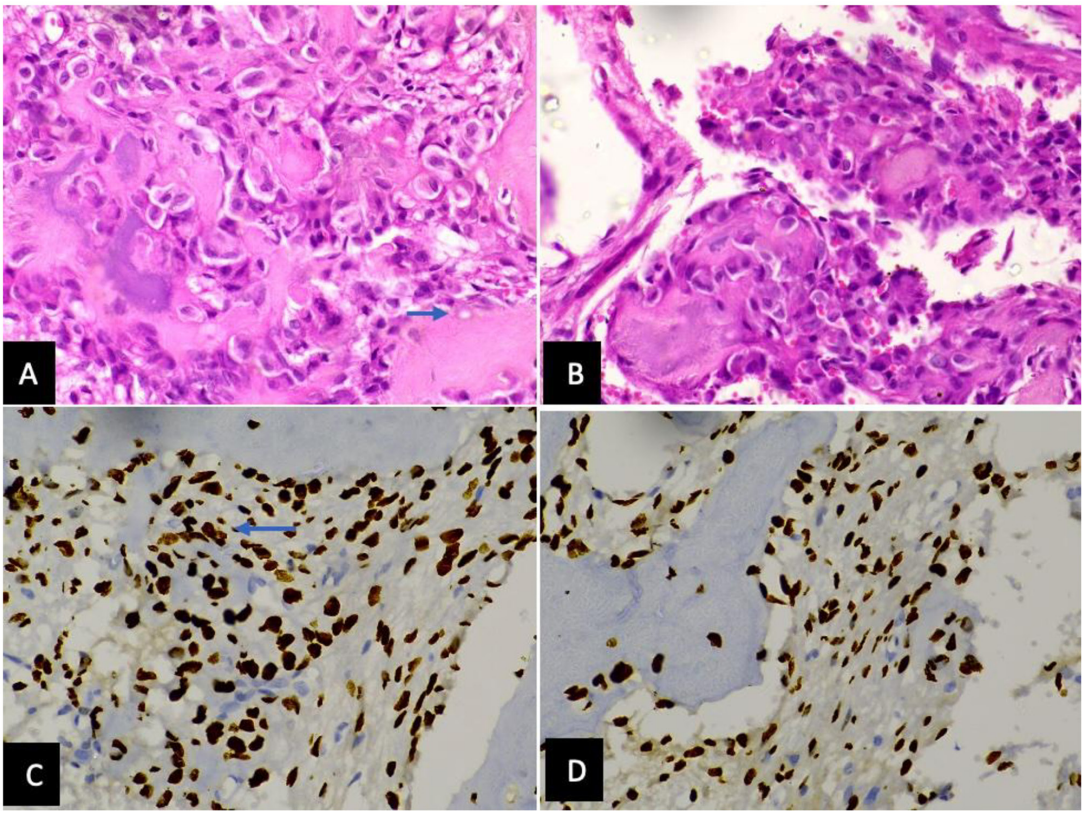

Figure 3 - (A) and (B) The histopathology report reveals increased vascularity in trabeculae of bone and focal areas of tumor osteoid with atypical cells showing features of hyperchromatism and pleomorphism resembling malignant osteoblasts. Few areas of chondrocytes, chondroid tissue and chronic inflammatory cells were also seen; (C) and (D) Immunohistochemistry is found positive for SATB2 in tumor cells.

radiographic findings, a provisional diagnosis of ossifying fibroma of the mandible, was given, and differential diagnosis of Myositis ossificans, osteomyelitis, osteoma, fibrous dysplasia, cement-osseous dysplasias, and OS [6] were considered. Incisional biopsy was performed, which revealed predominantly features of osteomyelitis, with increased vascularity in the trabeculae of bone and focal areas of tumour osteoid with atypical cells showing features of hyperchromatism and pleomorphism resembling malignant osteoblasts. A few areas of chondrocytes, chondroid tissue, and chronic inflammatory cells were also seen. Further, PETCT was taken, which revealed high uptake within a large expansile lesion centred over the right hemi mandible measuring 4.6 × 3.9 × 5.5 cm involving the body and ramus with few areas of cortical thinning and breaching. A perilesional node measuring 9 × 7 mm was also seen alongside bilateral FDG avid level lb largest measuring 1 × 0.7 cm on the left side, and an FDG avid hypodense lesion measuring 1.8 × 1.1 cm with centripetal enhancement was noted in the third segment of the left lobe of the liver, as shown in Figures 2D and 2E. On USG correlation, a corresponding irregular hyperechoic lesion was detected, suggestive of hemangioma, along with diffusely increased FDG uptake in the thyroid gland; no significant lesions were also noted. Simultaneously, immunohistochemistry (IHC) for SATB2 was done which revealed the fragments of lamellar bone with a foci showing neoplastic tumor cells with nuclear eosinophilic cytoplasm resembling malignant osteoblasts and presence of atypical cells riming the lace like osteoid. Adjacent fibrocollagenous tissue showed scattered chronic inflammation and congested blood vessels. Hence IHC for SATB2 revealed positive staining for tumour cells as shown in Figures 3C and 3D. Correlating the clinical, radiological and microscopic findings, features were suggestive of OS. Additional investigations, including chest radiographs and whole-body imaging, revealed no evidence of a metastatic tumour. The patient was referred to the department of Maxillofacial surgery for segmental mandibular resection. Free fibula flap was safely anastomosed to the superior thyroid artery with venuous drainage established through external and internal jugular veins. Postoperative recovery period was satisfactory. Patient is currently under follow up and is disease free since two years.

DISCUSSION

OS is an aggressive neoplasm that mainly affects the long bones and rarely affects the jaw bones. OS of jaw bones constitutes around 6-7% of overall OS and accounts for 0.2% of all malignancies [7]. Paget's disease, radiation therapy, existing bone pathology and genetic factors are some of the predisposing factors of OS. The maxilla and mandible are equally affected, with a slight predilection for the mandible. Posterior body and ramus are the most frequently affected, whereas in the maxilla alveolar ridge, floor of maxillary antrum, and the palatal bone of the maxilla are less commonly affected [8]. OS can present as an incidental radiographic finding with no symptoms or clinical signs [9]. However, the common symptoms include swelling at the diseased site followed by local pain, numbness, facial dysesthesia, mobility of teeth, trismus, limitation of jaw movements, headache and nasal obstruction or bleeding [10].

OS presents with varied radiographic appearances in the conventional radiographs, which include radiolucent, radiopaque and mixed radiographic presentations, including widening of the periodontal ligament space [11]. A case of OS reported in the maxillary antrum, demonstrating an extensive destructive lesion, also showed evidence of an asymmetric widening of the periodontal space [12]. The other classical presentations include radiopacity with the Codman Triangle, moth-eaten or sunburst appearance. When the neoplasm extends into the periosteum, irregular bony spicules project outward and perpendicular to the tumour, thereby giving the classical "sunburst" or "sunray" appearance [13]. Due to the diverse radiographic presentations, the pre-operative diagnosis may pose a challenge to the clinician. However, ill-defined, unicentric destructive lesions with sclerotic or lytic or mixed presentation should arouse suspicion for OS [14]. The present case showed ill-defined lytic regions in the right angle of the mandible in the orthopantomogram(OPG), thereby leading to the suspicion of OS. When compared with conventional radiographs, CT and MRI, FDG PETCT have been proven useful in the assessment of the extent of the tumour and for staging of the disease. PETCT has shown more advantages, especially in the accuracy of early diagnosis of OS, as it has demonstrated high sensitivity and specificity in the diagnosis of OS. Primary malignant bone tumours are characterised by the Warburg effect, leading to an increased glycolysis rate, thereby , higher uptake of the radiotracer in malignant cells. The hybrid imaging modality PETCT overcomes these limitations by fusing metabolic data with detailed anatomical information. This simultaneous assessment of molecular activity and morphology establishes PETCT as an efficient, single-modality tool for accurate whole-body staging and re-staging of patients with primary malignant bone tumours [15]. It provides information regarding the extent of primary lesion and presence of metastases, including response assessment to neoadjuvant chemotherapy and disease prognostication [16]. The non-characteristic initial presentation of OS often leads to misdiagnosis of OS as osteomyelitis (OM) . The present case showed a predominance of histopathological features in favour of OM; however, careful examination revealed a single focal area with osteoid and atypical cells, thus arousing a suspicion for a possibility of OS. As reported previously, with routine imaging not helping to differentiate between OM and OS, in the present case, PETCT proved to be a valuable diagnostic aid in arriving at a final diagnosis of OS. SATB2 is a proven IHC marker with a sensitivity and specificity of 90.4% and 95.3% respectively, in the detection of OS [17]. It has been a potent diagnostic marker in differentiating from other sarcomas mimicking OS, especially Ewings Sarcoma. However, since few high-grade chondrosarcomas express SATB2, it is recommended that the diagnosis of OS should be made correlating the clinical, radiological and histological features of malignant osteoid formation with positivity of SATB2. IHC plays a significant role in the identification of the origin of cells in both benign and malignant conditions, thereby proving to be a valuable adjunct in diagnosing challenging cases [18]. An aggressive OS was reported by Mohanavalli et al. [19] in the right side mandible with typical sunburst appearance in CT involving the symphysis and body of mandible. The patient was treated with neoadjuvant chemotherapy followed by wide local excision with reconstruction done using microvascular free fibula flap and titanium plate, similar to the present case [19]. Another similar case of mandibular OS reported by Alramadhan et al. [20] showing ill-defined destructive lesion with classical sunburst appearance in CT demonstrated SATB2 positivity in immunohistochemistry, similar to the expression shown in our present case. However none of the above-mentioned cases had PETCT done, which could be attributed to the straight forward radiographic appearance of extensive periosteal reaction [20]. The management protocol of OS includes chemotherapy and surgery. Chemotherapy is indicated for unresectable cases of head and neck OS, with distant metastases to the skull base or those presenting with aggressive behaviour in histopathology [21]. Currently, multimodality

treatment approaches by combining radical surgery, chemotherapy and/or radiation therapy has been frequently employed, resulting in better long-term prognosis and survival rates. OS of the jaw remain enigmatic, and a number of challenges are yet to be resolved in terms of diagnosis, as well as treatment and recurrence [22].

CONCLUSION

This case report highlights the crucial role of PETCT in the diagnostic workup of the initial presentation of OS. The addition of IHC with the routine histopathological examination can aid in the diagnosis of clinically challenging cases of OS, thereby helping in providing adequate treatment for better long-term prognosis and disease-free survival.

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Data availability

The datasets generated and/or analyzed during the current study are available from the corresponding author on reasonable request.

Author's Contributions

CVD: Conceptualization, Methodology, Investigation, Supervision, Writing – review & editing. DBG: Data curation, Formal analysis, Validation, Writing – review & editing. ES: Methodology, Investigation, Resources. LDJ: Methodology, Project Writing – review & editing. AW: Formal analysis, Software, Data interpretation. MN: Investigation, Writing – Original Draft Preparation.

Conflict of Interest

The authors declare no conflicts of interest.

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Regulatory Statement

A signed consent form was obtained from the patient to disclose the case.

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