Scholars Journal of Medical Case Reports
Abbreviated Key Title: Sch J Med Case Rep
ISSN 2347-9507 (Print) | ISSN 2347-6559 (Online)
Journal homepage: https://saspublishers.com

OPEN ACCESS

Oncology-Radiotherapy

Maxillary Osteosarcoma: A Rare Case Report and Literature Review

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DOI: https://doi.org/10.36347/sjmcr.2026.v14i01.002 | **Received:** 23.10.2025 | **Accepted:** 28.12.2025 | **Published:** 02.01.2026

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Abstract | **Case Report**

Osteosarcoma is a primary malignant bone tumor producing osteoid or bone. It is rare in the jaws, representing only 4% of all osteosarcomas, and maxillary osteosarcoma is even less common. This report presents a case of maxillary osteosarcoma in a 34-year-old woman. The patient presented with tingling, hypoesthesia of the right maxillary region, headaches, and moderate weight loss. Initial dental evaluation with panoramic radiograph revealed a maxillary lesion. Further imaging with MRI and CT showed an aggressive, heterogeneous lesion with calcifications, lysis of the orbital floor, and invasion of the nasal cavity. Diagnosis was confirmed by incisional biopsy, revealing a conventional osteoid-producing osteosarcoma. The patient underwent right hemimaxillectomy with tumor-free margins. Postoperative imaging revealed no residual tumor or metastasis. This case highlights that, despite the generally favorable biological behavior of jaw osteosarcomas, delayed diagnosis can lead to extensive local disease. Early recognition by dental and maxillofacial professionals is crucial for optimal management.

Keywords: Osteosarcoma, Maxilla, Bone Neoplasms, Diagnosis, Hemimaxillectomy, Rare Diseases.

INTRODUCTION

Osteosarcoma (OS) is the most common primary malignant tumor of bone [1]. However, when involving the craniofacial skeleton, it is considered a rare entity. Jaw osteosarcomas (JOS) account for only 4% of all reported osteosarcomas [1,2]. Histologically, they are characterized by an anaplasticstroma with direct production of osteoid matrix [3].

Although JOS and osteosarcomas of the long bones share similar histopathological features, JOS exhibits a distinct biological behavior [1,2,4–6]. It typically occurs 10 to 20 years later than osteosarcoma of the long bones and shows a lower tendency for metastasis. This may partly explain its more favorable prognosis; however, its initial presentation as a painless swelling can delaydiagnosis until an advanced stage [2,7].

The etiology of OS remains incompletely understood. Paget's disease, fibrous dysplasia, and exposure to ionizing radiation are recognized predisposing factors [2,4]. Increased cellular activity may also contribute to OS development, as the metaphyseal growth plate of growing bones represents the most commonly affected site [2].

We report a rare case of maxillary osteosarcoma in a 34-year-old woman, along with a review of the literature, aiming to better delineate the clinical, diagnostic, and therapeutic characteristics of this uncommon localization.

CLINICAL PRESENTATION

- **Patient identity and medical history**

The patient is a 34-year-old woman, married with no children, working as a pastry chef. Her relevant medical history includes type 2 diabetes mellitus managed by dietary measures alone and a previous surgery for an ingrown toenail. No drug allergies or toxic exposures were reported. Family history was unremarkable, with no similar cases identified.

- **History of the disease**

Over the past six months, the patient experienced tingling sensations in the right maxillary region, followed by localized anesthesia and headaches, in the context of a general health deterioration with unquantified weight loss.

She initially consulted a dentist, who performed a panoramic radiograph revealing a right maxillary lesion, and prescribed symptomatic treatment. Subsequently, she was referred to a neurologist who

requested a brain MRI. While no cerebral parenchymal or vascular abnormalities were detected, the imaging revealed an aggressive, heterogeneous, and calcified lesion of the right maxillary sinus, measuring 26 × 28 mm, associated with lysis of the orbital floor and invasion of the nasal cavity.

- **Additional investigations and diagnosis**

An incisional biopsy demonstrated a sarcomatous tumor proliferation expressing SOX9 and CD99, with a Ki-67 proliferation index of 30% and the presence of osteoid matrix, suggestive of a conventional osteosarcoma.

The diagnosis was confirmed following a right hemimaxillectomy with enucleation. Postoperative histopathological examination confirmed a conventional osteoid-producing osteosarcoma, with the tumor measuring 8 cm in its greatest dimension after reconstruction. Surgical margins were free of tumor, with no evidence of vascular emboli or perineural invasion.

- **Postoperative imaging**
  - **Facial CT scan:** Wide resection of the right maxilla with right orbital exenteration; no residual mass identified. Mild infiltration of the preseptal and jugal soft tissues was noted. The contralateral nasal cavities and sinuses were normal, and no pathological lymphadenopathy was observed.
  - **Thoraco-abdomino-pelvic CT scan (TAP CT):** No detectable secondary lesions, ruling out distant metastases.

- **Postoperative clinical examination**

The patient was conscious, with a World Health Organization (WHO) performance status of 1. The surgical scar was well healed, with no joint abnormalities or palpable lymphadenopathy.

CONCLUSION

This case involves a 34-year-old woman with type 2 diabetes mellitus, referred for further management of a high-grade right maxillary osteosarcoma following right hemimaxillectomy. Postoperative clinical evaluation was satisfactory, with preserved general condition (WHO performance status 1) and no evidence of metastatic disease on thoraco-abdomino-pelvic CT. The patient is currently under close postoperative surveillance and is scheduled to receive adjuvant chemotherapy based on methotrexate, along with radiotherapy.

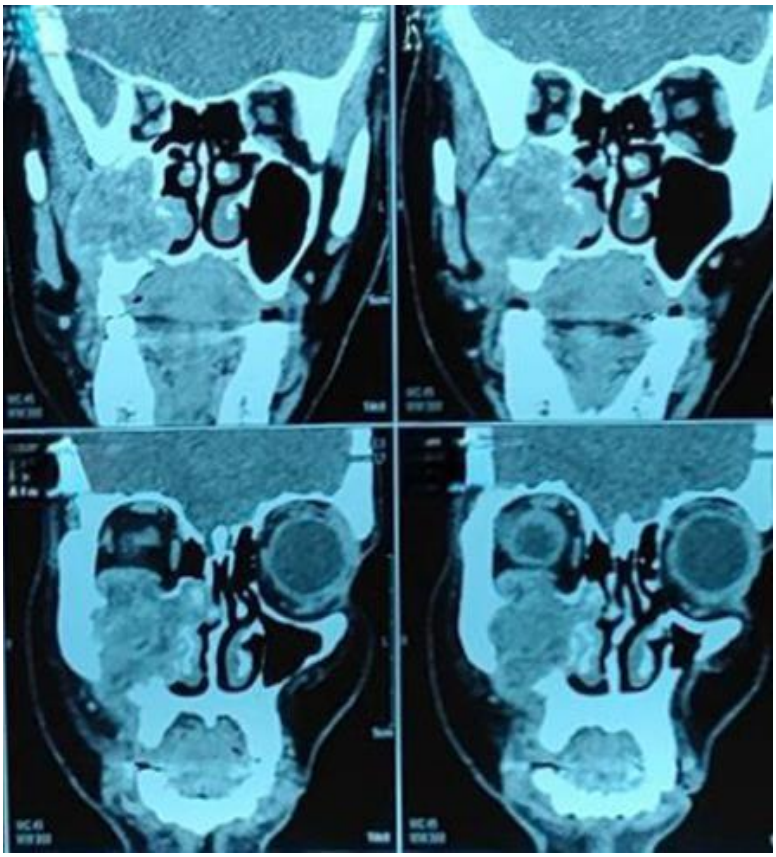

Figure 1: **Coronal section of the preoperative facial CT scan**

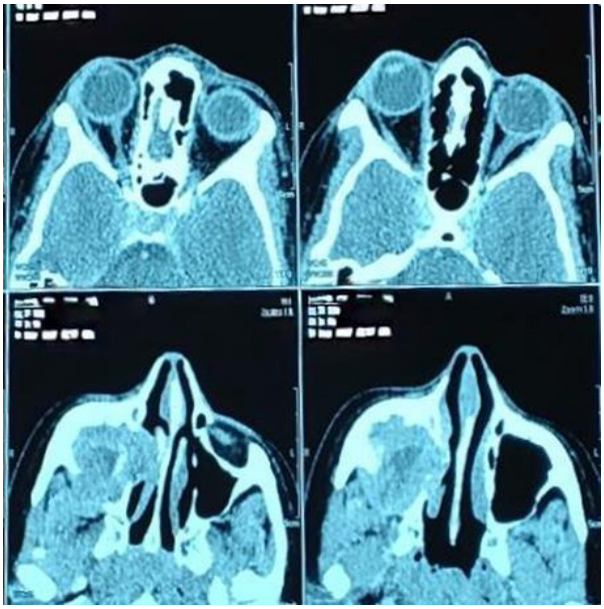

Figure 2: Axial section of the preoperative facial CT scan

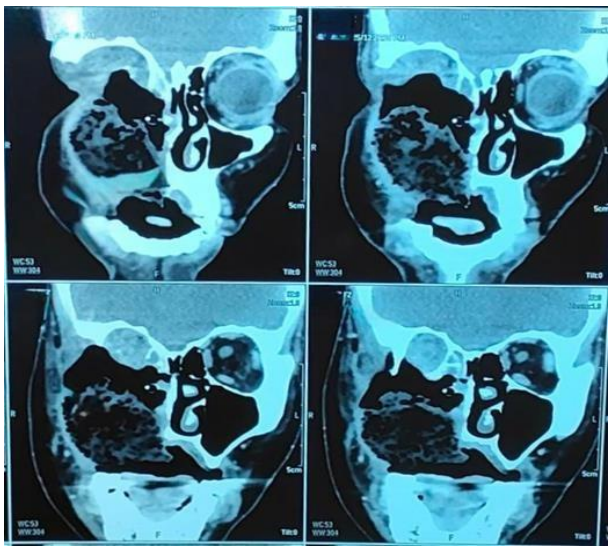

Figure 3: Coronal section of the postoperative facial CT scan

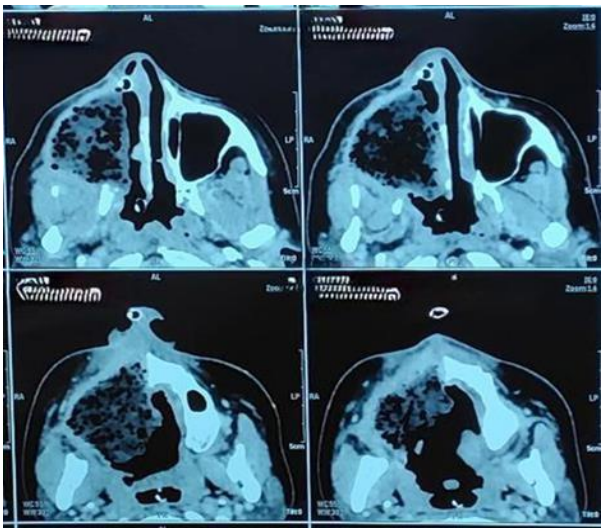

Figure 4: Axial section of the postoperative facial CT scan

DISCUSSION

Maxillary osteosarcoma (MOS) is a rare primary malignant bone tumor of the craniofacial region, accounting for only 4–10% of all osteosarcomas [8]. Unlike osteosarcoma of the long bones, which predominantly affects adolescents and young adults, MOS typically occurs in middle-aged adults, with a median age of approximately 35 years and a male-to-female ratio close to 1:1 [14]. In our case, the patient was 34 years old, which is consistent with the usual demographic profile.

The diagnosis of MOS relies on a combined clinical, radiological, and histopathological assessment [5]. Initial symptoms are often nonspecific, including tingling sensations, localized anesthesia, pain, or swelling, frequently leading to delayed referral to specialized care. In our patient, the initial manifestations consisted of tingling, right maxillary hypoesthesia, and headaches, followed bymoderate weight loss, illustrating the insidious onset commonly associated with this tumor.

Radiologically, MOS may present as mixed radiopaque and radiolucent lesions, with cortical bone destruction and characteristic periosteal reactions such as the “sunburst” appearance [17]. In our case, MRI and facial CT scans revealed an aggressive, heterogeneous lesion with calcifications, associated with lysis of the orbital floor and invasion of the nasal structures, highlighting the locally aggressive behavior typical of maxillary osteosarcomas.

Histopathological examination is essential to confirm the diagnosis. Conventional osteosarcomas are defined by the direct production of osteoid by an anaplastic stromal component. Depending on the predominant cell type, they may be classified as osteoblastic, achondroplastic, or fibroblastic [8]. In our patient, both the biopsy and postoperative pathological examination demonstrated a malignant osteoid-producing mesenchymal proliferation consistent with a conventional osteosarcoma, with tumor-free surgical margins and no evidence of vascular emboli or perineural invasion. Although the chondroblastic subtype is the most frequently encountered in jaw osteosarcomas, no clear correlation between histological subtype and prognosis has been established [20].

Complete surgical resection with negative margins remains the cornerstone of treatment and the main determinant of curability [25,26]. In the maxillary region, achieving adequate margins is often challenging due to the proximity of critical structures such as the orbit and the brain. Our patient underwent a right hemimaxillectomy with enucleation, followed by planning for adjuvant chemotherapy. Postoperative imaging, including facial CT and thoraco-abdomino-pelvic CT,demonstrated no residual tumor or distant metastases, confirming the effectiveness of surgical management and the absence of systemic spread.

The role of neoadjuvant or adjuvant chemotherapy in maxillary osteosarcomas remains controversial, in contrast to long bone osteosarcomas where it significantly improves survival outcomes [8,27–29]. In the present case, the indication for adjuvant chemotherapy was based on the high-grade nature of the tumor and the potential risk of local recurrence, despite the absence of metastatic disease.

Radiotherapy is generally considered to have limited efficacy and is usually reserved for unresectable tumors or cases with residual disease [8,30]. Overall, the prognosis of maxillary osteosarcomas is more favorable than that of osteosarcomas of the long bones, owing to their lower metastatic potential (approximately 18% versus 80%) and often intermediate-grade malignancy [8]. Nevertheless, diagnostic delay, as observed in our patient who initially consulted a dentist for seemingly benign symptoms, may result in extensive local disease, necessitating more radical surgery and adversely affecting function and aesthetics. This underscores the importance of heightened awareness among dental and maxillofacial professionals when confronted with persistent or atypical maxillary swellings.

CONCLUSION

This case highlights the typical yet misleading presentation of maxillary osteosarcoma, the critical importance of early diagnosis, the pivotal role of complete surgical resection with clear margins, and the consideration of adjuvant chemotherapy to optimize outcomes. Long-term clinical and radiological follow-up remains essential for the early detection of recurrence or postoperative complications.

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