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CASE REPORT

Osteosarcoma of the maxilla and maxillary sinus mimicking post-implant infectious complication: a case report

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Abstract:

Osteosarcoma is a rare malignant primary bone tumor in the gnathic bones and presents a locally aggressive behavior. Its diagnosis may be particularly challenging when it arises in uncommon sites such as the maxilla and maxillary sinus, especially in the presence of local conditions that simulate inflammatory or infectious processes. This study reports a case of high-grade osteosarcoma involving the maxilla and maxillary sinus in a female patient, initially misinterpreted as an infectious complication related to bone graft material extrusion following maxillary sinus floor elevation for dental implant placement. The patient presented delayed diagnosis and unfavorable clinical evolution, undergoing radical surgical resection followed by adjuvant radiotherapy and later palliative chemotherapy. This case highlights the diagnostic complexity of maxillofacial osteosarcomas and reinforces the importance of a multidisciplinary approach, careful interpretation of imaging findings, and early biopsy in atypical presentations to improve disease control and prognosis.

Keywords: Osteosarcoma; Maxilla; Maxillary sinus; Dental implants; Oral neoplasms.

INTRODUCTION

Osteosarcoma is a malignant primary bone neoplasm originating from mesenchymal cells capable of producing osteoid or immature bone. It is the most common primary bone sarcoma and predominantly affects the metaphyses of long bones, especially in adolescents and young adults. Although its etiology remains unclear, factors such as prior exposure to ionizing radiation, chemotherapy, and hereditary genetic syndromes are associated with increased risk[1-6].

Involvement of the gnathic bones and maxillary sinus is rare, accounting for approximately 6% of cases, with a higher prevalence in males, usually between the third and fifth decades of life. In this region, the mandible is more frequently affected than the maxilla[1-3]. Compared to long-bone osteosarcomas, gnathic tumors present a lower rate of distant metastases at diagnosis but demonstrate marked local aggressiveness, and primary lesions in this anatomical site are particularly uncommon[1,7,8].

Statement of Clinical Significance
Maxillary osteosarcoma can mimic post-implant infection, delaying diagnosis. Persistent or atypical symptoms after implant-related procedures should prompt comprehensive imaging, multidisciplinary evaluation, and timely biopsy to facilitate early diagnosis and appropriate management. This case emphasizes recognizing aggressive bone-forming lesions despite an apparent infectious context.

Histopathologically, osteosarcoma is characterized by malignant mesenchymal cells producing osteoid matrix, ranging from relatively uniform round or spindle cells to markedly pleomorphic cells with nuclear and cytoplasmic atypia. The amount of bone matrix varies considerably and may be associated with chondroid differentiation and fibrous connective tissue[1,5,6].

The mainstay of treatment is wide surgical resection with negative margins. Radiotherapy and chemotherapy may be used as adjuvant therapies in selected cases. In head and neck osteosarcomas, radiosensitivity

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is limited and the benefit of chemotherapy is less well established than in long-bone tumors, making complete surgical excision the most important prognostic factor. Overall five-year survival rates reported for gnathic osteosarcomas range from approximately 20% to 80%, depending on tumor site, stage, and margin status[1,2,4–7,9].

Clinically, the most common symptoms include pain, swelling, tooth mobility, and paresthesia. When the maxilla and maxillary sinus are involved, sinonasal symptoms such as nasal obstruction may occur[1-3]. Imaging plays a crucial role in diagnosis and treatment planning, with computed tomography (CT) and magnetic resonance imaging considered the gold standard for assessing tumor extension, cortical involvement, and mineralized components within soft tissues[2,5,6].

In implant dentistry, bone grafting associated with maxillary sinus floor elevation is widely performed. Among possible complications, graft material extrusion into the maxillary sinus and secondary infection may occur. The development of a malignant tumor in an area previously subjected to such procedures may represent an additional confounding factor, leading to diagnostic delay[10-18].

Therefore, this report aims to describe a diagnostically challenging case of osteosarcoma involving the maxilla and maxillary sinus, initially misinterpreted as an infectious complication related to bone graft material extrusion after dental implant placement.

CASE REPORT

A 62-year-old female patient was referred in 2025 to the Oral Medicine and Stomatology team of the Hospital das Clínicas, Ribeirão Preto Medical School, University of São Paulo (HCRP-USP), by the Otolaryngology service for joint evaluation. The referral was motivated by refractory acute sinusitis allegedly caused by extrusion of bone graft material following maxillary sinus floor elevation for dental implant placement in the right maxilla.

The patient reported dental implant placement in the right maxilla in 2015. Several years later, she developed recurrent infections in the implant region, initially managed with local measures by her dentist. In March 2024, due to persistent infection, the implants were removed. After removal, the patient developed progressive pain and swelling, unresponsive to antibiotic therapy, leading to referral to a tertiary care center with a presumptive diagnosis of odontogenic acute sinusitis.

Clinical examination revealed firm swelling of the right hemiface without signs of inflammation. Intraoral examination showed a firm swelling of the right hard palate, covered by intact mucosa with a focal ulcerated erythematous area, measuring approximately 4 cm at its greatest diameter (Figure 1). Adjacent teeth exhibited mobility, with positive pulp vitality tests. The patient did not report spontaneous pain or paresthesia at that time.

A CT scan performed outside the institution demonstrated partial occupation of the right maxillary sinus by an irregular, heterogeneous, predominantly hyperdense mass with multiple internal hyperattenuating foci suggestive of mineralized or calcified content. No evident cortical bone erosion or discontinuity was observed (Figure 2). The lesion was initially interpreted as extruded bone graft material from the previous sinus lift procedure.

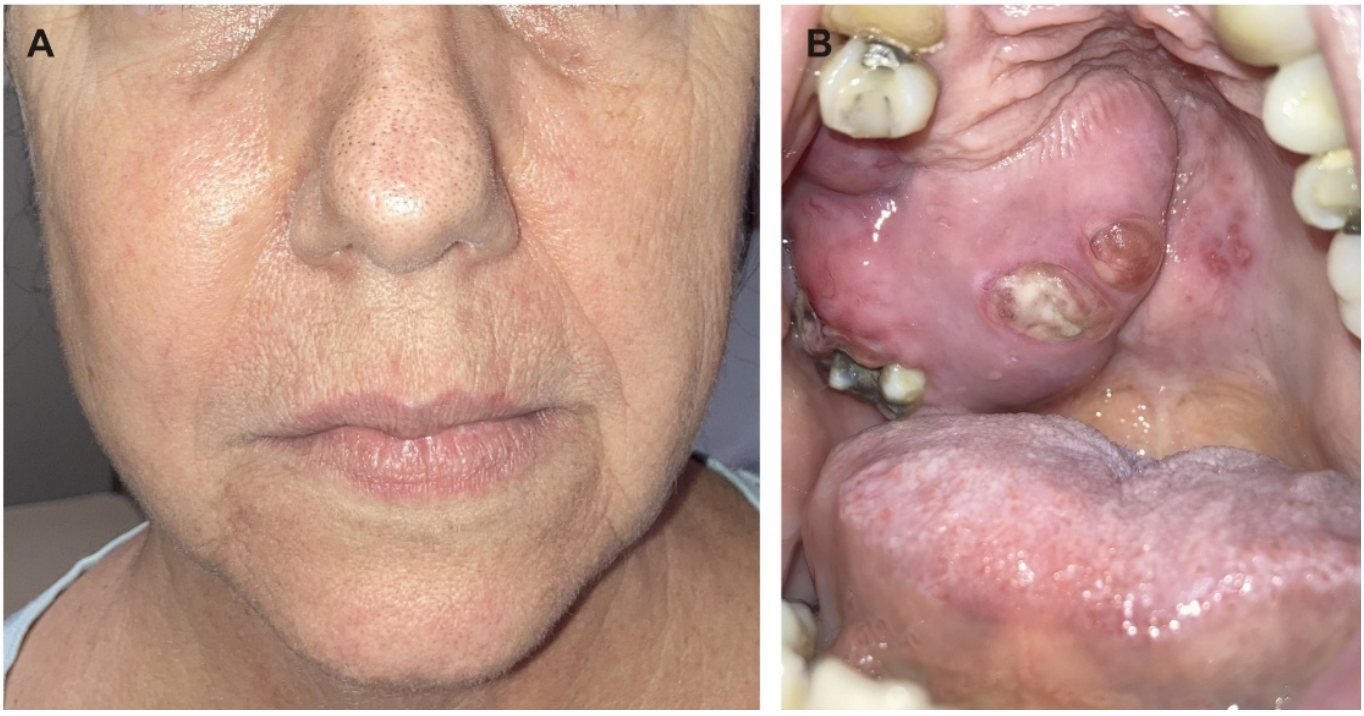
Previous imaging studies and clinical photographs were requested, including a CT scan obtained in March 2024 prior to implant removal (Figure 3). Due to persistent symptoms and atypical imaging findings, a malignant lesion was suspected. An incisional biopsy was performed following extraction of the most mobile tooth.

Histopathological analysis revealed proliferation of spindle-shaped malignant cells with enlarged hyperchromatic nuclei, frequent mitotic figures, areas of necrosis, and osteoid matrix formation with calcified areas. Immunohistochemistry showed diffuse positivity for CD99, focal positivity for SATB2 and smooth muscle actin (1A4), a high proliferative index (Ki-67 approximately 36%), and negativity for AE1/AE3, desmin, CD34, and S100 protein, consistent with high-grade osteosarcoma (Figure 4).

The patient underwent surgical treatment with sentinel lymph node evaluation by frozen section, which revealed malignant spindle-cell neoplasia with focal osteoid formation. Subsequently, right subtotal maxillectomy with modified radical neck dissection was performed. Reconstruction was achieved using a titanium mesh plate and a right temporalis muscle flap.

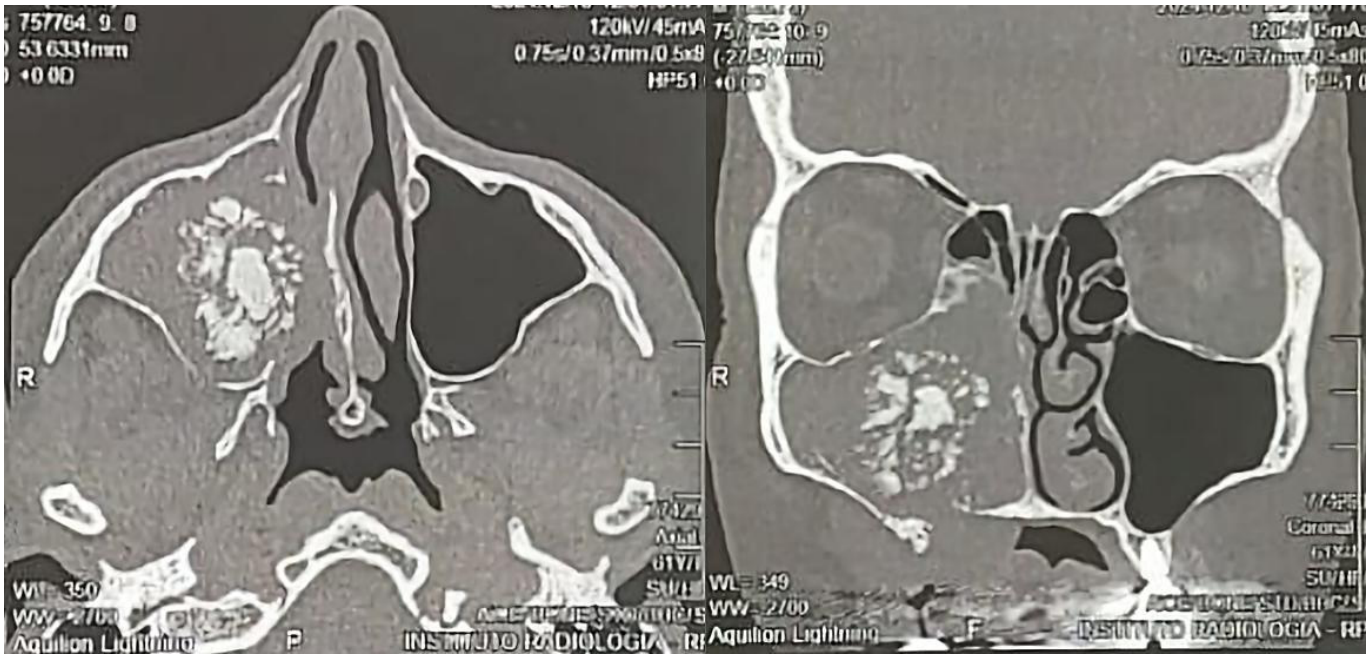
Final histopathology confirmed high-grade fibroblastic osteosarcoma measuring 5.5 cm, without angiolymphatic invasion, vascular invasion, or perineural infiltration. Among the 31 lymph nodes examined, only the sentinel lymph node was positive for metastatic disease. Focal involvement of the medial surgical margin was observed. Pathological staging was pT1 pN1 Mx.

Due to impaired renal function (creatinine clearance 45 mL/min), adjuvant chemotherapy was initially contraindicated, and exclusive radiotherapy was administered (70 Gy in 35 fractions). Four months after surgery, local disease progression and enlargement of pulmonary nodules were observed (Figure 5). Reduced-dose chemotherapy with doxorubicin and cisplatin was initiated, followed by second-line palliative chemotherapy



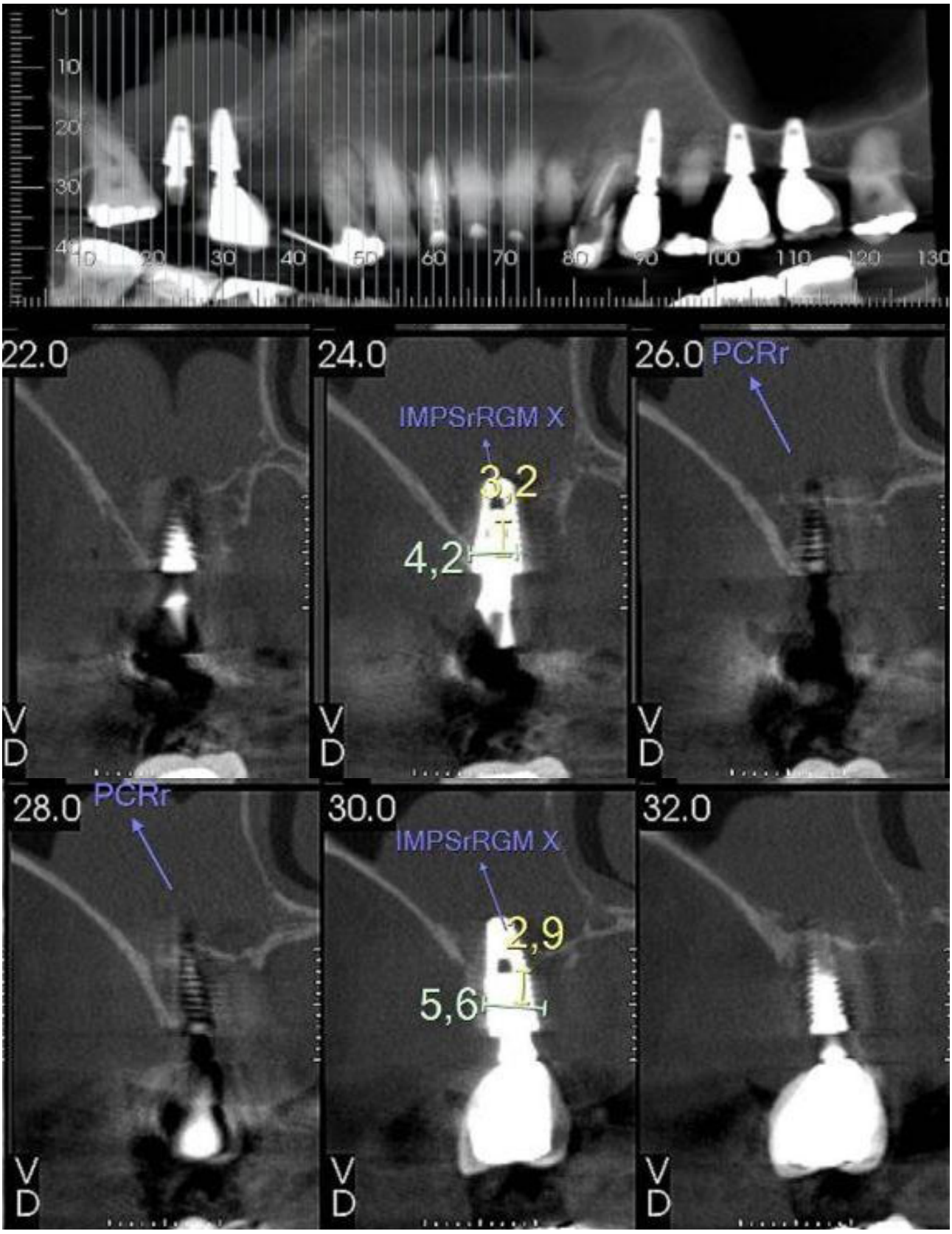
Source: Material from the authors themselves, obtained with the patient's authorization.

Figure 1. (**A**) Extraoral photograph showing right-sided facial swelling. (**B**) Intraoral photograph demonstrating a nodular lesion in the right palatal region.



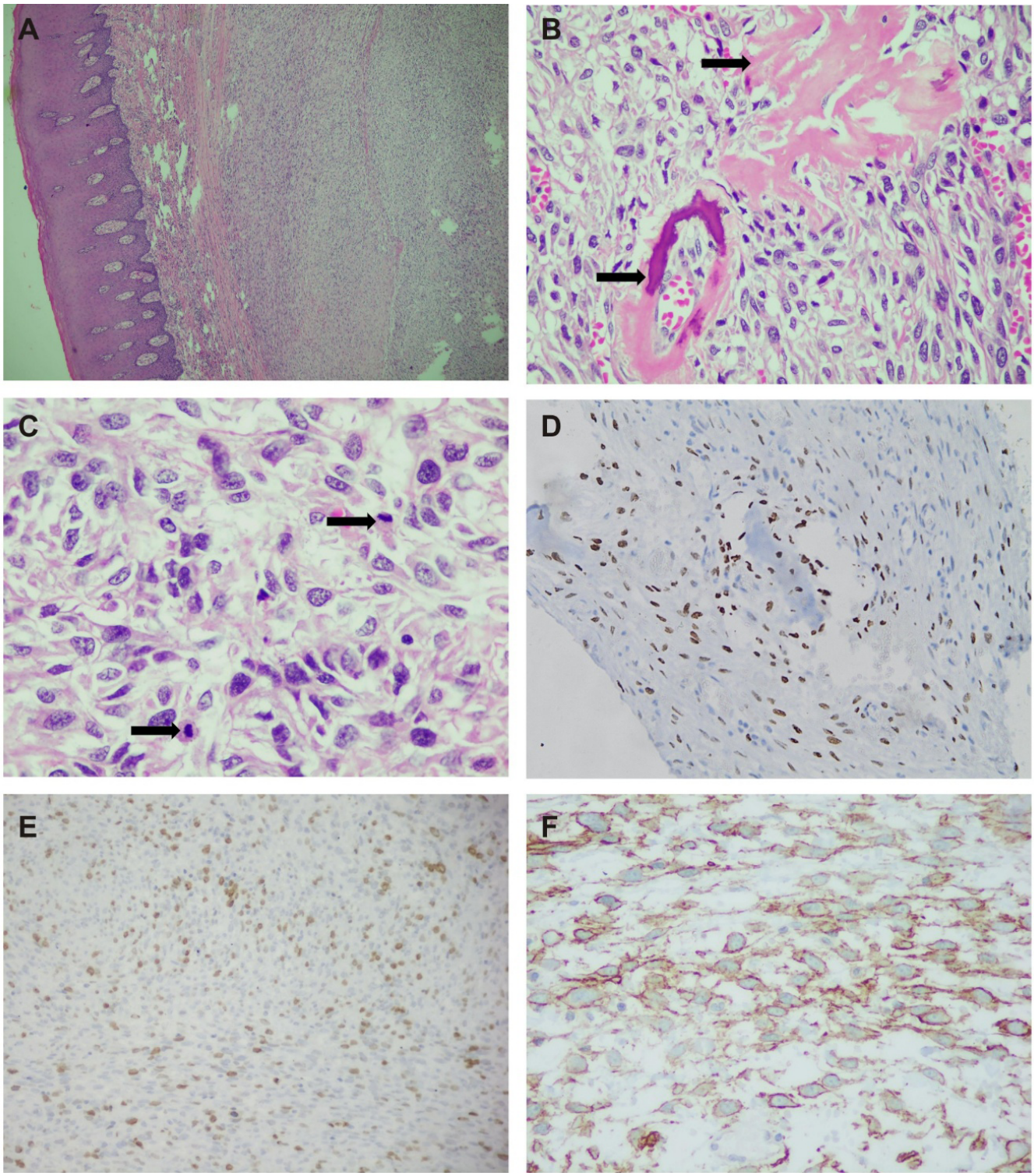
Source: Material obtained from the HCFMRP-USP imaging center, with the patient's authorization.

Figure 2. Computed tomography of the maxillary sinuses performed a few days prior to the tertiary care consultation.



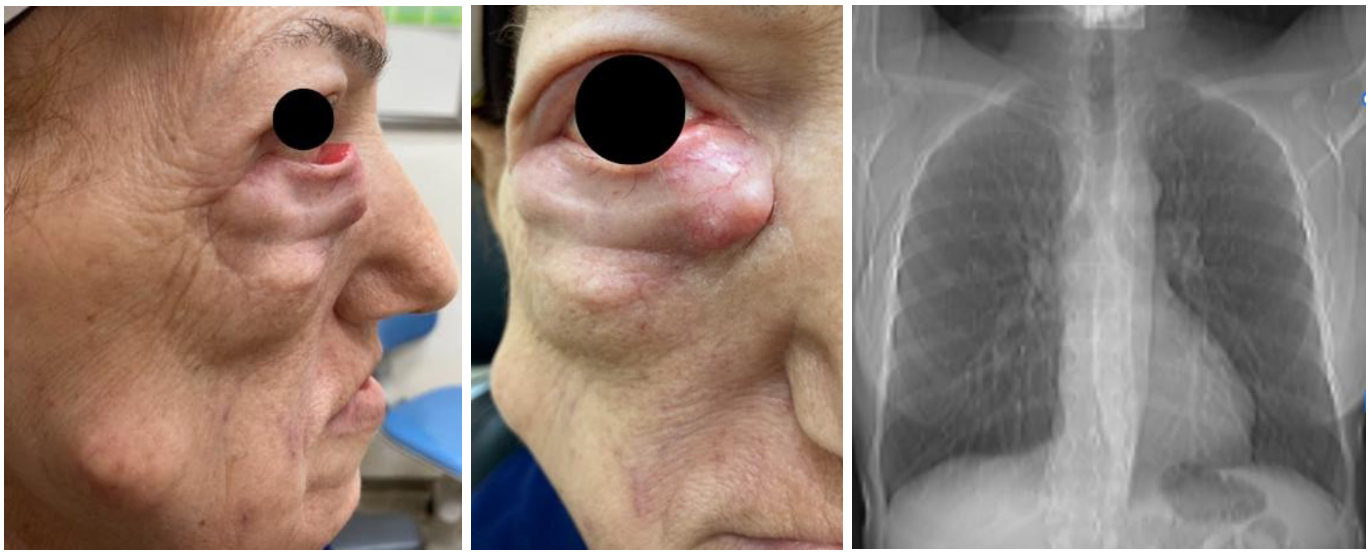
Source: Images provided by the patient.

Figure 3. Maxillary computed tomography requested by the dentist in March 2024. Note the limited visualization of the maxillary sinus.



Source: Images obtained from the Department of Pathology at HCFMRP-USP.

Figure 4. (**A**) Histopathological section showing the overall tumor architecture and the transition between the oral mucosa and the neoplastic lesion (hematoxylin and eosin stain, original magnification×40); (**B**) Osteoid production (arrows) by malignant neoplastic cells (hematoxylin and eosin stain, original magnification×400); (**C**) Malignant neoplastic cells exhibiting marked cellular atypia and atypical mitotic figures (hematoxylin and eosin stain, original magnification×400); (**D**) Immunohistochemical staining showing nuclear immunoreactivity for SATB2 in the neoplastic cells (original magnification×200); (**E**) Immunohistochemical staining showing nuclear immunoreactivity for Ki-67 in the neoplastic cells (original magnification×200); (**F**) Immunohistochemical staining showing membranous immunoreactivity for CD99 in the neoplastic cells (original magnification×400).



Source: Material provided by the authors, obtained with the patient's authorization. Radiographic image obtained at the HCFMRP-USP imaging center.
Figure 5. (**A, B**) Disease progression in the ipsilateral masseter and infraorbital regions. (**C**) Metastatic pulmonary nodules.

with ifosfamide and etoposide afterlocal recurrence and progression. Treatment was discontinued due to severe toxicity, and targeted therapy with sorafenib was introduced. The patient was transitioned to palliative care and died 15 months after surgery due to complications related to metastatic disease.

DISCUSSION

Gnathic osteosarcomas represent a small proportion of all osteosarcomas and typically affect male patients in the third to fifth decades of life[1]. The present case diverges from this epidemiological profile, involving an older female patient and affecting the maxilla, a less commonly involved site. Although maxillary osteosarcomas are less frequent than mandibular lesions, the main diagnostic challenge in the present case was not the anatomical location itself, but the extensive involvement of the maxillary sinus associated with clinical and radiographic findings that closely mimicked a post-implant infectious complication. This unusual presentation contributed to the initial diagnostic interpretation and delayed recognition of the underlying malignancy[1-6].

Immunohistochemistry plays a complementary role in the diagnosis of osteosarcoma, particularly in poorly differentiated tumors or when histopathological findings overlap with those of other spindle cell neoplasms. However, no immunohistochemical marker is sufficiently specific to establish the diagnosis independently, making correlation with clinical, radiographic, and histopathological findings essential. In the present case, immunohistochemistry was performed to confirm the histopathological diagnosis and to help exclude other spindle cell malignancies. SATB2 positivity supported osteoblastic differentiation, while the high Ki-67 proliferative index reinforced the high-grade biological behavior of the tumor. Although CD99 showed diffuse positivity, its limited specificity restricts its diagnostic value when interpreted in isolation. In contrast, the absence of AE1/AE3, S100, desmin, and CD34 expression helped exclude epithelial, neural, myogenic, and vascular neoplasms, thereby strengthening the diagnosis of high-grade osteosarcoma[1,6,11].

The understanding of the molecular biology of osteosarcoma has advanced significantly over the past decades. It is now recognized that this neoplasm is characterized by marked genomic instability, with frequent alterations involving the TP53 and RB1 genes, as well as dysregulation of pathways related to cell cycle control, DNA repair, and osteoblastic differentiation. Amplification of MDM2 and CDK4 is predominantly observed in low-grade osteosarcomas. Although these discoveries have substantially improved the understanding of osteosarcoma pathogenesis, their impact on routine diagnostic practice remains limited, and histopathological evaluation continues to be the cornerstone for the diagnosis of conventional osteosarcomas[6,19].

Despite demographic differences, the clinical presentation — facial and palatal swelling, pain, and tooth

mobility — was consistent with previously described manifestations of maxillary osteosarcomas. Sinonasal symptoms may further complicate diagnosis when the maxillary sinus is involved[1-6,11].

Surgical resection with negative margins remains the cornerstone of treatment. In contrast to long-bone osteosarcomas, the role of chemotherapy and radiotherapy in head and neck tumors is less clearly defined, and local disease control is the main determinant of prognosis[5,7]. Reported five-year survival rates vary widely and are strongly influenced by margin status and tumor extension[1,5-7,9].

In this case, diagnosis was delayed due to overlapping clinical and radiographic findings suggestive of post-implant infectious complications. The history of sinus floor elevation and the presence of mineralized material in the maxillary sinus contributed to initial misinterpretation. However, the presence of cortical bone destruction associated with an irregular heterogeneous mineralized mass occupying the maxillary sinus represented imaging features more consistent with an aggressive bone-forming neoplasm than with an isolated postoperative infectious process. The radiographic differential diagnosis should also include chronic osteomyelitis, fibro-osseous lesions, ossifying fibroma, chondrosarcoma, metastatic disease, and other primary malignant bone tumors[1-3,6,7,12]. Histopathologically, the differential diagnosis in the present case included sarcomatoid carcinoma, leiomyosarcoma, melanoma, malignant peripheral nerve sheath tumor, synovial sarcoma, and solitary fibrous tumor, all of which were excluded based on the combined histomorphological and immunohistochemical findings[1,6,11]. Limited imaging evaluation focused on the alveolar region and did not cover the maxillary sinus may have further delayed recognition of the neoplastic process.

The association between dental implants and osteosarcoma remains controversial and extremely rare, with only isolated case reports described in the literature. Although chronic inflammation and trauma have been proposed as potential contributors, no causal relationship has been scientifically established[6-12]. In the present case, implant therapy should be regarded as a confounding factor rather than an etiological agent.

This report emphasizes the need for comprehensive imaging, multidisciplinary evaluation, and early biopsy in patients with persistent or atypical complications following implant-related procedures involving the maxillary sinus.

CONCLUSION

This case highlights the diagnostic and therapeutic challenges of gnathic osteosarcoma, a rare and aggressive malignancy in which early diagnosis is critical for improved outcomes. Local factors related to dental procedures may obscure clinical presentation and delay diagnosis. Multidisciplinary assessment, appropriate imaging, and timely histopathological confirmation are essential for optimal management.

AUTHORS' CONTRIBUTIONS

TMI: Investigation, Writing – original draft. MAC: Methodology, Validation. MJP: Investigation. TCF: Validation. CANB: Investigation, Writing – review & editing. RBC: Resouces, Validation. GVC: Validation, Wirting – review & editing. HMAR: Validation, Writing – review & editing. ET: Investigation, Validation. LDM: Conception, Methodology, Supervision, Validation, Writing – review & editing.

CONFLICT OF INTEREST STATEMENT

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Ethics approval: Patient's written informed consent was obtained by the authors.

Declaration of generative AI and AI-assisted technologies in the writing process: During the preparation of this work the authors used ChatGPT in order to improve readability and language. After using this tool/service, the authors reviewed and edited the content as needed and take full responsibility for the content of the publication.

Data availability statement: The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

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