

CASE REPORT

Conventional osteosarcoma of the mandible: Report of a rare case

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Abstract

Osteosarcoma of the jaw can be successfully treated by a single aggressive surgical operation with good prognosis. Timely diagnosis, with a complete surgical removal of the tumor with clear margins, can fulfill favorable clinical results.

KEYWORDS

mandible, osteosarcoma, treatment

1 | INTRODUCTION

We are presenting a case of mandibular osteosarcoma in a 39-year-old, 13-week pregnant lady with a history of breast carcinoma. The tumor was successfully excised during a single surgical operation with uninvolved surgical margins. A two-year follow-up of the case revealed no evidence of disease recurrence.

Osteosarcoma occurs most commonly in long bones of the extremities near the metaphyseal growth plate. Osteogenic sarcoma of the jaw is a rare neoplasm, which represents about 7% of all OS and 1% of all malignancies in the head and neck region.^{1,2} Jaw osteosarcoma usually presents itself in the third and fourth decades of life, almost one to two-decade after their presentation in long bones; the neoplasm has a slight predilection for the mandible.³ The exact etiology of the disease is unknown. Three main factors may play essential roles in the development of the tumor, including irradiation, preexisting benign bone disorders, and genetic predisposition.^{1,4} Biologically, the OS of the jaw is considered

to be less aggressive, with a low incidence of metastasis and a better prognosis compared with those occurring in long bones.⁵ Gadwal et al. explained that the tumor size and resectability are two main prognostic criteria for the OS of the jaws.⁶ We report a case of mandibular osteosarcoma treated by surgical excision only in a pregnant female with a history of invasive ductal carcinoma of the left breast.

2 | CASE REPORT

A 39-year-old, 13-week pregnant lady was referred to an oral and maxillofacial surgeon for her facial asymmetry and painful swelling in the posterior mandible. Her past medical history included a history of invasive ductal carcinoma of the left breast, which was treated by left radical mastectomy and chemotherapy about three years ago. Intra-oral examination of the patient showed swelling of the right posterior mandible with intact mucosal surface and bony-hard consistency (Figure 1).

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FIGURE 1 Clinical view of the lesion. A mandibular expansile lesion with intact surface mucosa and bony-hard consistency

Based on the patient's history, the first differential diagnosis was distant metastasis of the previous malignancy. The X-Ray findings of the mandible were suggestive of an ill-defined mixed lytic lesion resorbing the roots of the

regional teeth (in the panoramic view) (Figure 2A). Further radiological investigations using Magnetic Resonance Imaging (MRI) of the patient's mandible revealed a large hypersignal lesion with ragged borders, cortical bone perforation and dental root resorption (Figure 2B). These radiographic features would also support a diagnosis of osteosarcoma of the jaw.

An incisional biopsy of the tumor was performed for definitive diagnosis. A malignant neoplasm composed of spindled to epithelioid cells admixed with extensive areas of amorphic eosinophilic materials mimicking osteoid formation. Also, "lace-like" osteoid formation was evident in some areas, which is compatible with conventional intramedullary osteosarcoma (Figure 3). In view of the histopathologic grade of the tumor (no necrosis and few mitotic features) and the patient's clinical situation (13-week pregnant), surgical treatment of the tumor involving the whole excision of the lesion without chemotherapy was considered as the best treatment option. The lesion was totally excised with uninvolved, clear margins (a least 1.5 cm) by partial mandibulectomy (Figure 4). The definitive diagnosis after evaluation of the subserial section of the lesion confirmed intramedullary osteosarcoma. The greatest dimension of the lesion was 5.5 centimeters. Her child was born full-term through an elective caesarian section in a healthy situation. We followed up the patient in our post-operation clinic for two years and have not found any clinical evidence of recurrence of the tumor, and the patient has no complaint.

3 | DISCUSSION

Osteosarcomas constitute about 40%–60% of all primary malignancies of the bones.^{1,7} Although the osteosarcoma of the jaw is a sporadic tumor, it is the most common malignant neoplasm of the jaws, subsequent to hematopoietic malignancies.⁸

The maxilla and mandibular bones are affected equally, but there are studies that suggest a slight predilection of



FIGURE 2 Radiographic features of the lesion. (A) Panoramic view shows a lytic ill-defined radiolucent-opaque lesion with irregular resorption of the mandibular first molar tooth roots. (B) A coronal view of the MRI of the patient. Large hypersignal lesion in the right mandibular body

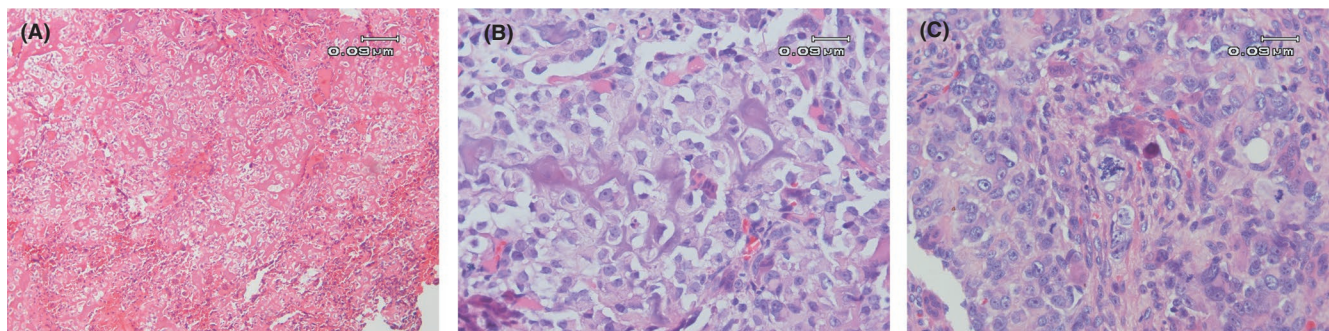


FIGURE 3 Histopathologic views. (A) Bone forming tumor with plump epithelioid type cells in the hemorrhagic background. (B) Plump osteoblastic cells with lace-like osteoid formation. (C) Numerous atypical mitotic figures and multinucleated giant cells

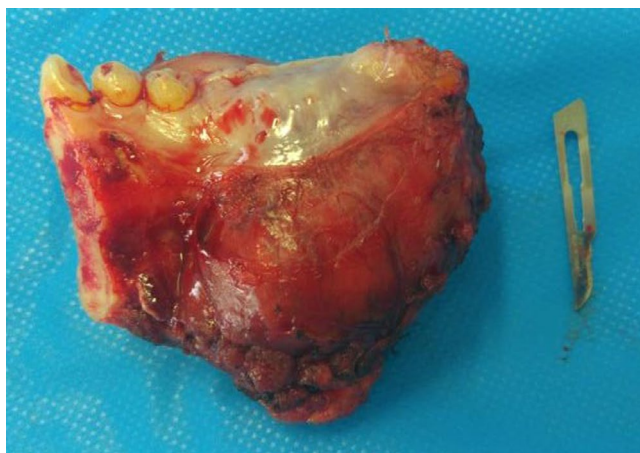


FIGURE 4 Gross specimen of mandibular surgical resection

the tumor for the mandible.^{1,3} In general, jaw osteosarcoma affects males more commonly than females and with an average age of 33–36 years old. The tumor presents in the jaw about one to two decades later than osteosarcoma of the long bones.^{5,8} To our knowledge, the case we are presenting is a rare one in terms of involving a female during pregnancy.

The most common signs of gnathic osteosarcoma are pain and swelling, but paresthesia and ulceration may also be present.⁹ Elkordy et al., in a case series study, reported that ulceration of the tumor is more common in the central lesions more than in lesions.³

As with other bone tumors, radiological findings are of significant importance in developing a differential diagnosis. The bone resorption and bone-formation nature of osteosarcomas lead to a mixed appearance of osteogenic and osteolytic patterns in the bone, reflecting in radiological studies.¹⁰

Although computer tomography (CT) is commonly used for evaluation of jaw pathologies, but it seems that MRI modality is a necessary tool to assess the exact extension of the tumor, especially in osteosarcoma of the mandible, in which evaluation of the mandibular canal involvement is essential.^{3,11}

In our reported case, both panoramic X-Ray imaging and MRI findings were in favor of the diagnosis of the osteosarcoma identified by an ill-defined mixed osteolytic lesion and its local influence on the regional structures such as irregular resorption of dental roots.

Histopathological features of osteosarcoma can present with a broad spectrum. For instance, conventional intramedullary osteosarcoma of the long bones is composed of the proliferation of neoplastic mesenchymal cells with spindle-shaped, epithelioid, or plasmacytoid appearance. The immature osteoid formation produced by malignant lesional cells is the histological key to diagnosis.¹

Osteoblastic and chondroblastic subtypes of the osteosarcoma are the most common histological groups of gnathic osteosarcoma.^{12–14} Aggressive surgical resection and advanced reconstruction techniques are the most critical factor in the treatment of jaw osteosarcoma. Particular attention needs to be paid to achieve clear surgical margins that play a crucial role in the treatment and the prognosis of osteosarcoma.¹⁵ The plan for the surgical treatment and the applied technique depends on the possibility of an effective and functional reconstructive surgery.¹⁶

The biological behavior of the osteosarcoma of the jaws is very different from long-bone tumors. Gnathic osteosarcoma is more likely to recur locally, with a lower rate of distant metastasis in comparison to osteosarcoma of long bones.^{1,16} We reported mandibular osteosarcoma in a 39 years old pregnant woman who was treated by a single surgical operation.

Many factors can influence the overall prognosis of gnathic osteosarcoma, including the patient's age, tumor size, histopathological grade response to chemotherapy, and tumor resectability at the time of presentation (1, 2, and 6). The clear and uninvolved surgical margins of the excised tumor, as achieved, seems to be the main prognostic factor in our case. Mandibular osteosarcomas possess a better prognosis than maxillary cases, which can also explain the favorable clinical outcome in our case.^{3,17} It may be due to better access and

easier resection of mandibular lesions in comparison with maxillary cases.

In patients who had their tumors removed surgically and the resected margins are involved by tumoral cells, multimodality therapy for cancer should be considered. In the cases of inadequate response to chemotherapy, further studies are needed. However, radical resection is the main factor for treatment and cure in jaw osteosarcomas.¹ Mortality in patients with osteosarcoma lesions is rare, and in case of death, the cause is a local extension of the tumor to the brain.¹⁸ In this case, we have demonstrated that complete surgical excision of gnathic osteosarcoma with uninvolved resected margins could cure the patients without introducing systemic treatment modalities.

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CONFLICT OF INTEREST

There is no conflict of interest.

AUTHOR CONTRIBUTIONS

(1) Hassan Mirmohammad Sadeghi contributed to patient treatment. (2) Abbas Karimi contributed to patient treatment and data collection. (3) Samira Derakhshan contributed to preparation of the manuscript and pathologic report. (4) Pouyan Aminishakib contributed to pathologic report. (5) Kiarash Parchami contributed to final preparation of the manuscript, data collection, and submission.

ETHICAL APPROVAL

This paper was approved by “Tehran University of Medical Sciences.”

CONSENT

Patient consent form is signed by the patient.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

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