

Osteosarcoma of the mandible in a older woman: a case report

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Abstract:

Osteosarcoma is a malignant tumor with mesenchymal origin, most commonly found in extremities, pelvis and axial skeleton, including only 6% with mandibular and maxillary involvement. In jaws, this lesion is, generally, a single lesion with predilection for posterior ramus mandible and men in the third or fourth decade of life. The aim of this study is report a 57-year old woman diagnosed with osteosarcoma in the posterior mandible region treated with surgical resection associated to bone and soft tissue reconstructive techniques.

Keywords: Osteosarcoma. mandible. Women

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INTRODUCTION

Osteosarcoma is the most common malignant mesenchymal tumor that affects the bones, especially the long bones and, more rarely, the jaws. Approximately 6% of all cases affect the jaws. This tumor produces immature and osteoid bone, which proliferates through cell stroma.¹

Tumor etiology remains uncertain, but some factors seem to contribute to the risk of its development, such as the male gender, third and the fourth decade of life, genetic pattern, pre-existing bone changes and trauma in the area.² Both the maxilla and the mandible can be affected, with a slight predilection for the ramus of the mandible.¹

Radiographically, osteosarcoma shows radiolucent with ill-defined edges or mixed appearance, both radiopaque and radiolucent. Cortical destruction and periosteal reaction perpendicular to the cortical bone are also observed, giving the appearance of “sun rays”.³

The World Health Organization (WHO) classification of bone tumors recognizes several variants that are histologically classified into different histologic subtypes. The type of extracellular matrix produced by the neoplastic cells determines the classification of osteosarcomas, including conventional, telangiectatic, small-cell, low-grade central, secondary, periosteal, parosteal, and high-grade surface. The most frequent type is the conventional osteosarcoma, which is classified into three subtypes: osteoblastic, chondroblastic, and fibroblastic osteosarcoma^{4,5}. Histologically, there is a proliferation of atypical, ovoid or spindle cells, which produce an osteoid matrix, immature bone and chondroid matrix^{5,6}

The recommended treatment in the jaws is aggressive surgical resection, with a wide safety margin. Chemotherapy / radiotherapy is an adjunct to treatment.^{2,7} It is important to highlight the need for reconstruction of the surgical area, using grafts and flaps. The prognosis is better when osteosarcoma has not reached large proportions and complete surgical removal is possible with negative margins. Osteosarcoma in gnathic bones has a superior prognosis than in long bones, but the number of recurrences is relatively higher.⁸

The aim of this study is report a case of the osteosarcoma in the mandible in a 57 year old woman that was treated with surgical resection associated to reconstruction techniques.

CASE REPORT

A 57-year-old woman patient was referred to the Dentistry Department of Pontifical Catholic University of Minas Gerais, Brazil, with historic of burning mouth and lips, for 6 months.

At the first consultation, in the extra oral examination, no noticeable changes were observed. The intra oral examination revealed a slight elevation covered by erythematous mucosa in the posterior right side of the buccal region of the mandible, with moderate pain on palpation. (Figure 1) Complementary radiographic and tomographic examinations were performed.



Figure 1. Intraoral appearance showing an expansive and diffuse swelling of the mandible with the erythematous aspect of the overlying mucosa

Panoramic radiography showed a poorly delimited radiolucent area of bone destruction in the right posterior region of the mandible, which mimics a osteomyelitis appearance. (Figure 2) In the initial Computed tomography (CT) the report was of a benign fibro- osseous lesion. An incisional biopsy was performed, and the material was sent for anatomopathological analysis. (Fig. 3A and Fig B). Histological examination showed proliferation of atypical osteoblast-like cells, with cellular and nuclear pleomorphism, hyperchromatic nuclei, atypical mitosis and osteoid formation. (Fig. 4A and Fig B).

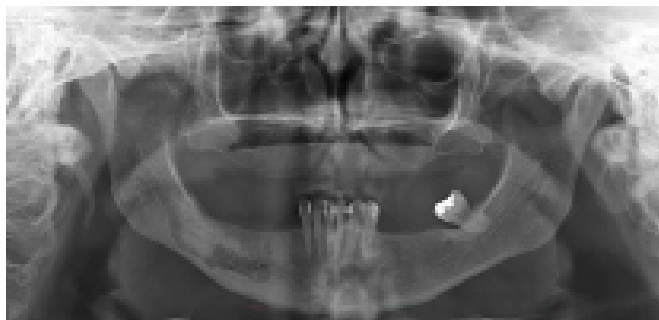


Figure 2. Panoramic radiography showing bone reabsorption poorly defined, with radiopaque areas, in the right mandible body and angle

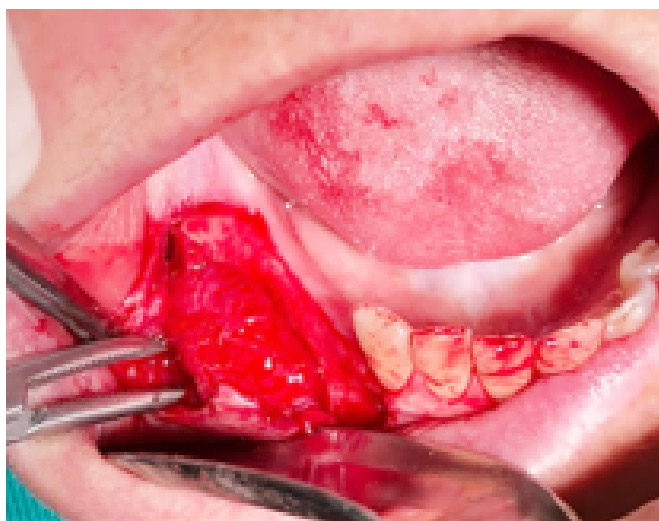


Figure 3. A) Surgery approach to perform the incisional biopsy of the lesion; B) Proximity of the lesion with the mental nerve

The diagnosis of osteosarcoma was established, and the patient was referred for treatment in an Oncology Service. Unfortunately, four months later the patient had not received any treatment for the osteosarcoma. The tumor, at this time, presented worrying changes with great increase in volume inducing facial asymmetry. (Figure 5)

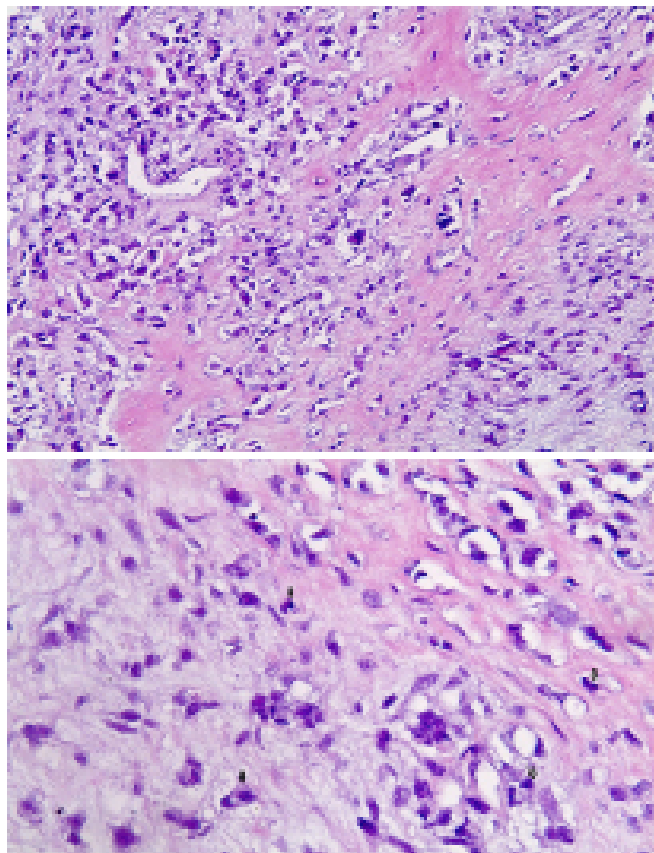


Figure 4. A) Proliferation of atypical spindle cells in sheet patterns and formation of immature bone trabeculae; B) Magnified view showing detail of the cellular atypia



Figure 5. Frontal view of the swelling inducing facial asymmetry, four months after diagnosis

The intra oral aspect now was of marked expansion on the posterior side of the mandible, measuring approximately 40 millimeters in length, extending from the body of the mandible until the angle of mandible, with an erythematous appearance of the lining mucosa. In the face of clinical and imaging aspects of the lesion, the main diagnostic hypothesis was osteosarcoma.

A second CT was performed where a mass hyperdense, with ill-defined limits in the region of the mandibular body and mandibular ramus on the right side, associated with hypodense areas of great medullary cortical osteolysis, which infiltrated into the soft tissue, closely related to the mandibular canal and foramen mental, showing an area of bone neoformation, with aspects of "sun rays", involving the buccal and lingual cortical, was observed. The tomography report was of osteosarcoma (Fig. 6). Six months after the diagnosis the patient received surgical treatment of the tumor. The medical approach was partial surgical resection, with reconstruction of the region with non vascular bone from the fibula associated to pectoralis muscle major flap. However, a year later, there was a tumor recurrence with involvement of the left side of the mandible of the patient. Due to difficulty in hospitalization because of the COVID-19 pandemic the patient is waiting for a new surgical opportunity and chemotherapy treatment was started, as the logical and possible option in the face of this contingency.

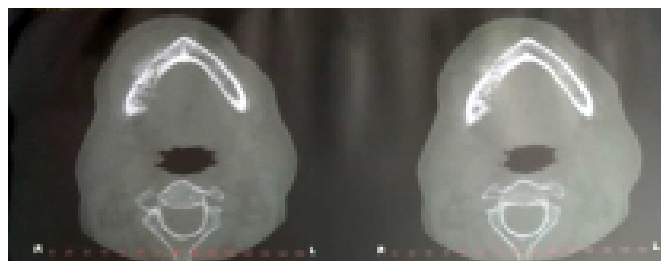


Figure 6. Computed tomography in axial sections-hyperdense show mass, with ill-defined limits in the mandibular body and ramus on the right side, associated with hypodense areas of great cortical spinal destruction and bone neoformation, with aspects of sun rays

DISCUSSION

Osteosarcoma is a malignant tumor with mesenchymal origin, most commonly found in extremities, pelvis and axial skeleton. When localized in the craniomaxillofacial region comprise less than 10% of all cases of osteosarcoma, with involvement of the mandible and the maxilla accounting for 6%. Osteosarcoma is the second most common malignant bone tumor in this region, only after the bone marrow tumors like multiple myeloma.^{1,9,10}

In jaws this lesion is more frequent in the posterior ramus of the mandible, being, commonly, presented as a solitary lesion.^{5,6,10} In this present case, the lesion was localized in the body and angle of mandible.

This disease usually affect persons in the third or fourth decade of life, with mean age between 33 and 36 years., with individuals being diagnosed 10-20 years older than the patients with this lesion in other parts of the body.^{5,11} Osteosarcoma of the jaws show more predilection for men compared to women, with the ration of 1.4:1.⁷ The osteosarcoma reported here differs from the scientific literature, since it affected a woman in her sixth decade of life.

The etiology of osteosarcomas remain unknown, but some predisposing factors have been proposed. This factors include intraosseous diseases (fibrous dysplasia or cemento-ossifying fibroma), Li-Fraumeni syndrome, previous radiotherapy in the region, Paget disease, multiple osteochondromatosis, chronic osteomyelitis, trauma, like tooth extraction, myositis ossificans, molecular alterations and mutation in tumor supressor genes p53 and RB1.^{4,5,11,12}

The osteosarcoma spreads along bone marrow spaces and mandibular canal. The extraosseous spread is facilitated by inferior alveolar nerve at the mandibular foramen, periodontal ligaments and mental nerve.¹¹ Swelling is the most common sign, but pain, paresthesia, ulceration, loosening or separation of teeth are related in some cases.^{1,2,5} The initial patient complaint was burning mouth and lips, but in the intra oral exam, was observed the swelling in the posterior region of the right side of the mandible.

This finding led us to make the imagiologic exams that suggesting a neoplastic intraosseous lesion compatible with an osteosarcoma, which was confirmed through histopathological analysis of excised in incisional biopsy. After three months, the tumor showed great growth, leading to facial asymmetry.

Generally, this tumor represent an ill-defined and destructive lesion, with features of osteogenesis (sun ray effect), osteolysis or a combination of both, depending of stage of osteosarcoma. The other radiologic features include periodontal ligament widening, lamina dura attenuation around a tumor.^{1,5,6} Although the radiologic exam is an auxiliary tool in diagnosis, the histopathologic features is the gold standard in diagnostic.¹⁰ The radiologic description related in this case report was a ill-defined and osteolytic image associated to areas of bone neoformation, with a typical sun ray appearance.

In the histological analysis, the features of the osteosarcoma is composed by chondroblastic, osteoblastic or fibroblastic types, with presence of nuclear atypia, atypical mitosis and anaplastic stromal cells.^{2,5,10} The malignancies features as the described above were found in this present case, however without chondroblastic aspects.

After adequate diagnosis, the main treatment of osteosarcoma of jaws is resection with wide margins (greater than 5 mm), but this modality in these regions can be difficult due to proximity to the vital structures (alveolar inferior nerve). In cases of positive surgical margins, the adjuvant radiotherapy improve the outcomes and local control.^{1,2,5,9,13} In relation to chemotherapy, there are not many studies that improve the effectiveness in management of this lesion in facial region, due to the richer vascularity, which may auxiliary on recurrence of the tumor cell after chemotherapy.^{10,13} The modality of treatment chosen for this woman was resection with absence of positive margins, without the use of radiotherapy and chemotherapy at the first moment.

The prognosis of this pathology in the jaws is better than in long bones, due to a lesser incidence of metastasis, with a rate of 18% and occurring in a later stage, most commonly found 2 years after initial diagnosis.^{1,11} The most favorable prognosis is evident in younger patients, radical surgery with negative margins, small tumors (less than 6 cm), absence of soft tissue involvement and early diagnosis. In contrast, the local recurrence varies between 17 to 70% for osteosarcomas in the jaws, being more significant than these lesions in the extremities, which present a rate varying between 5 to 7%. The survival rates of osteosarcomas of the jaws is about 77% at 5 years.^{2,11,12,14} Unfortunately, after one year of the surgical treatment, our patient developed local recurrence of the osteosarcoma. The recommended treatment was adjuvant chemotherapy and a new surgical intervention. The behavior of this tumor can affect the woman prognosis.

According Yutaro et al, 2017, although no consensus exists regarding the efficacy of chemotherapy in head and neck osteosarcoma, a few studies have shown that chemotherapy in neoadjuvant and adjuvant setting improves survival in patients with head and neck osteosarcoma. However, the main prognostic factor in osteosarcomas of the head and neck region still remains the radical surgery involving complete tumor resection with negative margins.⁴

Although the resection is the modality of choice for treatment of osteosarcomas, this technique generates a bone defect, disruption of muscular attachments and nervous injury, interfering in mastication, swallowing, speech, protection of airway and facial aesthetics.^{2,5} To improve these conditions, can be used reconstruction plate, flaps, bone grafts and a combination of these options.^{5,15,16,17} In this present case, reconstruction was made using a non vascular fibula graft associated to pectoralis muscle major flap. Free flaps are the gold standard in the mandibular bone reconstruction. Although this, the non vascular graft can be used for defects up to 9 cm, being a reasonable option for mandibular reconstruction, including iliac crest, fibula, calvarium, rib and tibia as the most common donor sites. The fibula is a good option due to the possibility of two-team approach, availability of 20–25 cm of bone for harvest and least donor site morbidity. The combination of this graft with pectoralis major flap aids in the adequate soft tissue closure and protection of the bone graft, avoiding the exposure and infection, being a good option, especially, for patients with chronic comorbidities and low socioeconomic condition.^{16,17,18,19}

CONCLUSION

It is important to highlight the difference in tumoral behavior of osteosarcoma of the jaws and its behavior in long bones. Sometimes osteosarcoma mimics other oral lesions like inflammatory osteomyelitis and periapical cysts, requiring differential diagnosis between them. Early diagnosis is essential for a better prognosis, since surgical resection of the tumor with negative margins is the best treatment, which allows for less mutilating surgery. At the moment, there is an ongoing debate about the value of neoadjuvant or adjuvant chemotherapy in the management of head and neck osteosarcomas. However more studies will be necessary to clarify the clinical impact of chemotherapy in the conduct for treatment of the jaw osteosarcoma.

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