

Fast Growing Jaw Osteosarcoma in a 55 Year Old Woman-A Case Report

Abstract

Background: Osteosarcoma is an osteoid or bone producer malignant tumour. Fast growing bones are frequently affected like metaphysical growth plates in the femur. Jaw Osteosarcoma is very rare, with an incidence of 0.7 per million, and have not been extensively evaluated.

Case presentation: A 55-years-old woman presented tooth mobility and a dental extraction was realized, with volume increase in the area next four weeks. After another four weeks of medical treatment without volume reduction biopsy was developed and histopathology report mandibular osteosarcoma. She was evaluated by oncology surgery and left hemi mandible disarticulation was planned a week later. After that week tumour progressed 70% in volume, crossing the midline, causing inability to eat and respiratory distress. It was treated initially with chemotherapy, with a 60% volume reduction and programmed for hemi mandibulectomy after chemotherapy cycles conclusion. Unfortunately the patient died after the 4th chemotherapy cycle.

Conclusion: Osteosarcoma is a very rare entity with an aggressive presentation that must be suspected and diagnosed promptly because the high growing rate could predispose bone extension, face deformation, airway compromise and dissemination. Early identification and management with chemotherapy and radiotherapy preoperatively could achieve better outcomes.

Keywords: Jaw osteosarcoma; Bone malignant tumors; Cytoreduction

Abbreviations: JOS: Jaw Osteosarcoma; OS: Osteosarcoma; CT: Computed Tomography; CHT: Chemotherapy; RT: Radiotherapy

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Introduction

Osteosarcomas are rare bone tumours with presence of malignant mesenchymal cells producing osteoid or immature bone. They account for 40% - 60% of malignant bone tumours and presented a fast growing rate [1]. The peak incidence is in the fourth decade of life, and males are affected more frequently than females. Jaws Osteosarcoma (JOS) is extremely rare, representing about 7% of all osteosarcoma and 1% of all head and neck malignant tumours [1, 2]. Although it is comparatively rare and more frequent in long bones, osteosarcoma is still a common primary bone tumour of the jaw [3]. Clinical manifestations in the oral cavity include pain, swelling, displacement and loss of associated teeth, spasms, paraesthesia and nasal obstruction [4]. Treatment of JOS should be approached with radical surgery, although it cannot be contemplated as the sole treatment. Perioperative chemotherapy and radiotherapy must be included to achieve better results [5, 6].

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Case Presentation

A 55 year old female with no medical background had a dental piece extraction secondary to tooth mobility in the left lower jaw, followed by an increase in volume and loss of sensation in the left chin the next 4 weeks. After 4 weeks of medical treatment without volume reduction she was sent to maxillofacial surgery and a biopsy was done, reporting mesenchymal malignant neoplasia compatible with mandibular osteosarcoma (**Figure 1**). On physical examination an evident facial asymmetry secondary to left jaw tumour was observed, tumour expansion produce ipsilateral nasal obstruction and nasobuccal purulent exudate, left hemimandible loss of sensibility, impossibility to mouth close and tongue displacement to the right without airway obstruction, no cervical nodes were palpated (**Figure 2**). A head and neck CT scan was performed to evaluate resectability and reported: bone tumour from left lower jaw body, measuring $80 \times 76 \times 33$ millimetres which destroys the cortical and grows in latero-cranial direction, having an ovoid shape. Neck Zone II ganglia of 7 mm to 14 mm are observed. She was sent to oncology surgery department for left hemimandible disarticulation but the tumour had significant progression in a week increasing volume 70%, crossing the midline causing inability to eat and respiratory distress. Urgent tracheostomy and gastrostomy were performed (**Figure 3**). Chemotherapy was started for cytoreduction and 20% volume reduction was obtained with one cycle (**Figure 4**). After three more cycles a 60% decrease in volume was observed (**Figure 5**). Unfortunately the patient died after 4th chemotherapy cycle.

Discussion

JOS represents only 7% of all Osteosarcomas (OS) and occurs more frequently in long bones. Only 10% of the OS occurs in the head or neck region, and the most frequently affected bones are the maxilla and the mandible [1]. The peak OS incidence is between

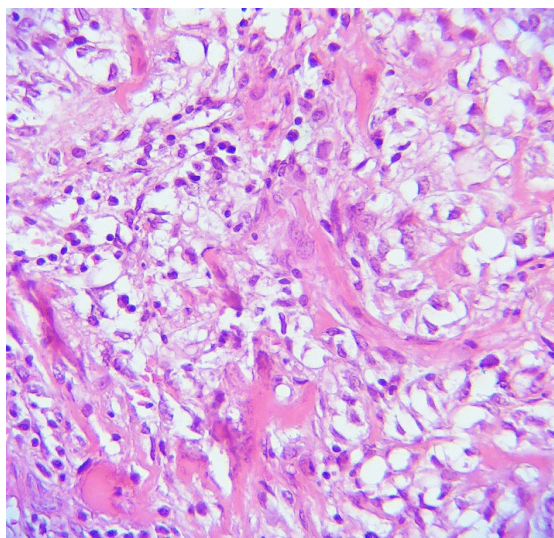


Figure 1 40x haematoxylin-eosin microscopy showing mesenchymal malignancy of pleomorphic cells with pleomorphic nuclei, 1-2 nucleoli of histiocytoid aspect that individually produce osteoid material.

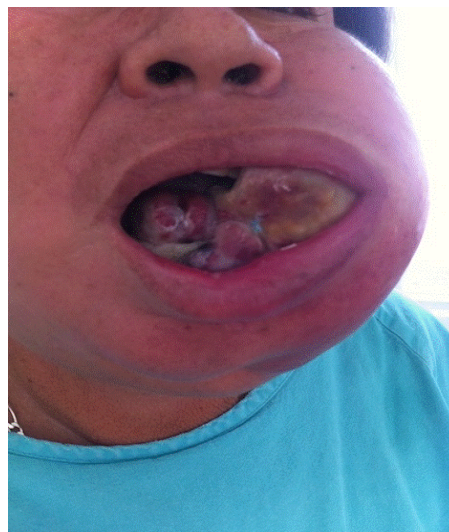


Figure 2 Patient 8 weeks after dental extraction. Jaw tumour producing facial asymmetry, impossibility to mouth closure and tongue displacement to the right.



Figure 3 Patient 9 weeks after dental extraction. Increase of 70% of tumour volume, crossing the midline and conditioning inability to eat and respiratory distress.

10 and 14 years old and is present in long bones. In the cases of JOS they are usually diagnosed two decades later. The most common signs and symptoms associated with this neoplasm consist in local swelling, mobile teeth, persistent pain and paresthesia. The diagnosis should always include panoramic radiographs, CT scan of head and neck to evaluate involvement of cortical bone and associated lymphadenopathies [2]. Other imaging studies should include magnetic resonance to evaluate the extent of the OS into the bone marrow. In order to stage the tumour we should rule out metastasis to other bones and organs so the chest and abdominal CT scan and isotope bone scan is needed [2-4]. For early diagnosis and preclusion of misdiagnosis as a benign lesion the pathologist should evaluate the patient clinical status [3]. The



Figure 4 1st Chemotherapy cycle with 20% decrease in volume and superficial necrosis zones.



Figure 5 4th Chemotherapy cycle presenting a 60% tumor decrease allowing oral feeding.

histological type of three major subtypes can be distinguished by their histological characteristics: osteoblastic, chondroblastic and fibroblastic, and two other rare subtypes: telangiectatic and small cell OS [3, 4]. In contrast to their counterparts the OS affecting the head and neck have a lower tendency to recur locally and are associated with 20% - 30% lower metastatic rate [2, 7]. The treatment of sarcomas will be different accordingly to location, histopathological type, metastatic disease and tumour size. The objective in the treatment is surgical tumor eradication, reconstruction and ultimately blocks resection with immediate reconstruction [5].

Surgery is the angular stone in treatment and the key factor to modify the prognosis of the patient. Based in a wide resection, clear one centimetre margins were associated with improved survival by 70%, which is not simple to achieve due to the complex anatomy of the maxillofacial region [5-8]. Also, multimodal treatment including pre or postoperative chemotherapy (CHT) is focused in cases of high grade tumours, positive surgical margins, lesions larger than 5 centimetres and unresectable lesions like in the presented case, factor that delay an urgent resection. Combined treatment with surgery and chemotherapy provides an 80% survival rate at 5 years [6]. Perioperative CHT provides an important benefit in JOS treatment, because it involves less destructive surgery and increases the opportunity to obtain clear margins, eradicate micro metastases and improve detection of tumour chemo sensitivity for better local and distance control [6-11]. Radiotherapy (RT) role in the multimodal treatment of JOS are not clear yet, nevertheless it is recommended in cases of distant metastases and positive margins [7-11]. Prognosis depends on different factors including histological subtype, malignancy grade, tumour size, patient age and chemotherapy response [5-8].

Conclusion

JOS is a highly malignant tumour, a very rare entity with an aggressive presentation that must be suspected and diagnosed promptly because the high growing rate could predispose bone extension, face deformation, airway compromise and dissemination. Early identification and management with chemotherapy and radiotherapy preoperatively could achieve better outcomes.

Ethical Considerations

Informed consent for publication of this case and photographs was signed. No conflict of interest in the presentation of this case.

References

- 1 Harazono Y, Yamashiro M, Yoshitake H, Kayamori K (2015) A case of highly suspected small cell osteosarcoma in the mandible. *Journal of Oral and Maxillofacial Surgery, Medicine and Pathology* 27: 38-40.
- 2 Wang S, Shi H, Yu Q (2012) Osteosarcoma of the Jaws: Demographic and CT Imaging Features. *Dentomaxillofacial Radiology* 41: 37-42.
- 3 Tabatabae SH, Jahanshahi G, Marvasti FD (2015) Diagnostic Challenges of Low-Grade Central Osteosarcoma of Jaw: a Literature Review. *J Dent Shiraz Univ Med Sci* 16: 62-67.
- 4 Angiero F, Moltrasio F, Cattoretti G, Valente MG (2011) Clinical and Histopathological Profile of Primary or Secondary Osteosarcoma of the Jaws. *Anticancer research* 31: 4485-4490.
- 5 Lecornu MG, Chuang SK, Kaban LB, August M (2011) Osteosarcoma of the Jaws: Factors Influencing Prognosis. *J Oral Maxillofac Surg* 69: 2368-2375.
- 6 Ferrari D, Codecà C, Battisti N, Broglio F, Crepaldi F, et al. (2014) Multimodality treatment of osteosarcoma of the jaw: a single institution experience. *Med Oncol* 31: 171.
- 7 Baumhoer D, Brunner P, Castori SE, Smida J, Nathrath M, et al. (2014) Osteosarcomas of the jaws differ from their peripheral counterparts and require a distinct treatment approach, Experiences from the DOESAK Registry. *Oral oncology* 50: 147-153.
- 8 Kämmerer PW, Shabazfar N, Makoie NV, Moergel M, Al-Nawas B (2012) Clinical, therapeutic and prognostic features of osteosarcoma of the jaws-Experience of 36 cases. *Journal of Cranio-Maxillo-Facial Surgery* 40: 541-548.
- 9 Dunfee BL, Sakai O, Pistey R, Gohel A (2006) Radiologic and Pathologic Characteristics of Benign and Malignant Lesions of the Mandible. *Radiographics* 26:1751-1768.
- 10 Thariat J, Julieron M, Bouchet A, Italiano A, Schouman T, et al. (2012) Osteosarcomas of the mandible: Are they different from other tumor sites? *Critical Reviews in Oncology / Hematology* 82: 280-295.
- 11 Thariat J, Schouman T, Bouchet A, Sarini J, Miller RC, et al. (2013) Osteosarcomas of the mandible: multidisciplinary management of a rare tumor of the young adult a cooperative study of the GSF-GETO, Rare Cancer Network, GETTEC / REFCOR and SFCE. *Annals of Oncology* 24: 824-831.