International Journal of Otorhinolaryngology and Head and Neck Surgery
Babu H et al. Int J Otorhinolaryngol Head Neck Surg. 2021 Apr;7(4):698-700
http://www.ijorl.com
pISSN 2454-5929 | eISSN 2454-5937

DOI: https://dx.doi.org/10.18203/issn.2454-5929.ijohns20211199

Case Report

A case of recurrent chondroblastic osteosarcoma mandible

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Received: 25 January 2021
Accepted: 03 March 2021

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ABSTRACT

Osteosarcoma is an uncommon primary malignant tumor of bone. Craniofacial osteosarcomas constitute only about 6.5–7% of all osteosarcomas. The most common histopathologic type is chondroblastic type in head and neck group and osteoblastic in extremity group. We present a case report of 21 year old male patient with chondroblastic osteosarcoma with two episodes of recurrence. The tumour was dealt with left hemimandibulectomy followed by radiochemotherapy. After first recurrence he underwent right hemimandibulectomy. During second recurrence excision of growth followed by radiochemotherapy was done. Thus an aggressive multi-modality approach was adopted for treatment. Osteosarcoma is an uncommon primary malignant tumor of bone. Craniofacial Osteosarcomas are considered a separate category in view of their low histologic grade, less frequent metastases and better prognosis. The most common presentation is local swelling with or without pain. Aggressive surgical approach with post-surgical radiochemotherapy can be an effective tool.

Keywords: Chondroblastic osteosarcoma, Osteosarcoma, Mandible, Hemimandibulectomy

INTRODUCTION

Osteosarcoma is an uncommon primary malignant tumor of bone and comprises of a group of rare primary malignant neoplasms of bone which exhibit considerable variation both in the clinical and histological appearance but also in the course and prognosis of the disease.[1]

CASE REPORT

Craniofacial osteosarcomas are considered a separate category in view of their low histologic grade, less frequent metastases and better prognosis. Hence the diagnosis of this variant is important.[2] The most common histopathologic type is chondroblastic type in head and neck group and osteoblastic in extremity group. Tumor-positive margin exists more frequently in head and neck OS, and local recurrence was the most common treatment failure on account of probable technical difficulty to achieve clear margins because of delicate and complicated anatomy.[3]

Therefore, the aim of our article is to make the clinician aware of this rare clinical presentation, treatment methods adopted and histopathologic challenges encountered.

A 21 year old male patient presented with complaints of progressive painless swelling left side mandible size 10×7 cm. There was no history of previous exposure to radiation or any surgery. On contrast enhanced computed tomography (CECT) intramedullary lytic lesion observed with presence of Codman's triangle along the outer cortex of mandible. Biopsy was done and diagnosed as osteosarcoma of mandible. Patient underwent left segmental hemimandibulectomy with left free fibular graft reconstruction in February 2017 (Figure 1). All margins were negative for malignancy. Histologic type was chondroblastic osteosarcoma. Post-surgery he underwent radiotherapy (30 fractions, 60 gray). Adjuvant

chemotherapy of 6 cycles with cisplatin and doxorubicin was given.

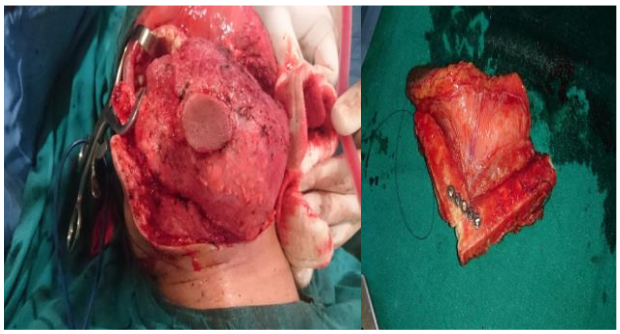

Figure 1: Intra-operative photograph showing the tumour and left fibular graft for reconstruction.

He was asymptomatic for 1 year but developed right mandibular swelling size 7×5 cm. CECT was done and diffuse infiltrative lesion involving right mandible with periosteal elevation was noted. In December 2018 he underwent right segmental hemimandibulectomy with right free fibular flap reconstruction (Figure 2). All margins were free of malignancy and histologic type was chondroblastic osteosarcoma (Figure 3).

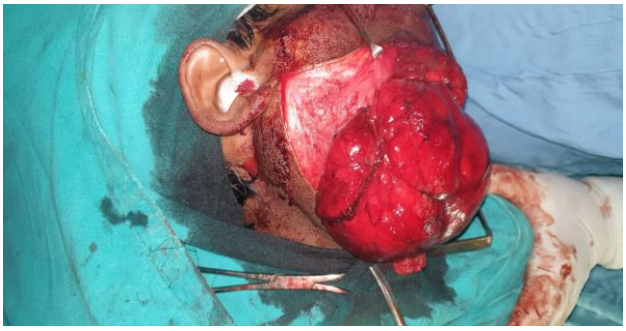

Figure 2: Osteosarcoma involving right half of mandible (recurrence).

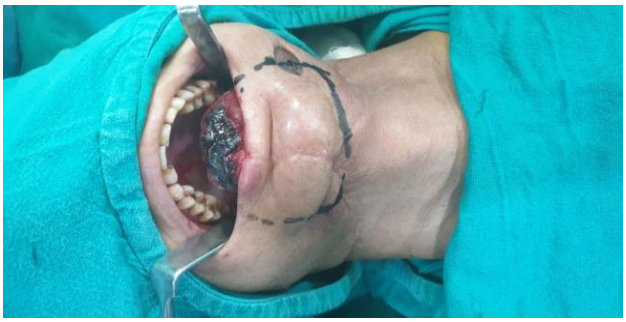

Figure 3: Tumour involving the mid mandibular region (recurrence).

After 3 months patient further developed swelling in the junction between left and right fibular graft. The growth was removed with repair of defect by PMMC flap in March 2019. Highly pleomorphic cells with coarse chromatin and irregular nuclear membrane were present. Final diagnosis was high grade variant of osteosarcoma. Patient underwent radiotherapy (30 fractions, 54 gray) and is currently undergoing chemotherapy-cisplatin and doxorubicin. The patient is under continued follow-up.

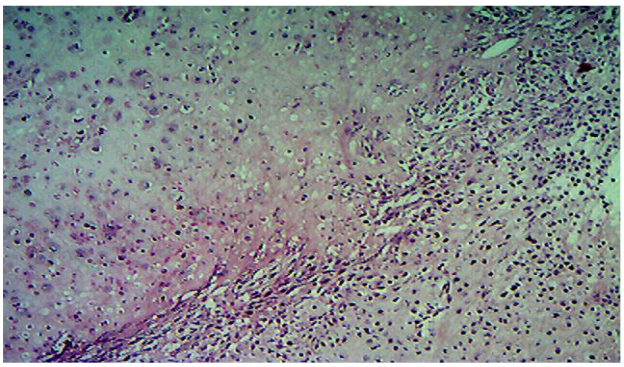

Figure 4: cartilaginous differentiation with tumour cells present in lacunar spaces (H&E 10x).

DISCUSSION

Chondroblastic osteosarcoma (CO) is a variant of osteosarcoma which are the most common malignant tumours of the skeleton.It most commonly occurs in the long bones of the extremities near metaphysical growth plates.[4] The classic osteosarcoma is a rare (0.2% of all malignant tumours) highly malignant tumour. It has been described in other bones including mandible, clavicle, cuboid and pelvis but in lower frequency.[5] Craniofacial osteosarcomas, are considered a separate category in view of their low histologic grade, less frequent metastases and better prognosis. Hence the diagnosis of this variant is important.[2]

The most common presentation is local swelling with or without pain.[6] In our patient also the presentation was swelling in the mandibular region. Our patient had no predisposing factors such as previous radiation exposure, fibrous dysplasia, Paget's bone disease.[7] Osteosarcoma may show various histopathological patterns. This creates problems in correct diagnosis hence histopathological examination of the fragments from all parts must always be done as the type of osteosarcoma may affect the treatment and the prognosis. In our patient absence of chondroid material in the second recurrence further supports this fact.[8]

Wide surgical resection is the primary treatment modality for jaw bone osteosarcoma, but obtaining clear surgical margins is difficult because of anatomic constraints. The highly aggressive nature of tumour predisposed to recurrences despite clear surgical margins.

Some studies show adjuvant chemotherapy may improve early survival, but neither chemotherapy nor radiotherapy appears to impact the long-term survival. Positive surgical margins are associated with poor prognosis, few authors have shown that the use of multimodal therapy may

significantly reduce rates of local recurrence, and thereby improve survival.[9] In our patient tumour was dealt with left hemimandibulectomy followed by radiochemotherapy. After first recurrence he underwent right hemimandibulectomy. During second recurrence excision of growth followed by radiochemotherapy was done. Thus an aggressive multimodality approach was adopted.

CONCLUSION

Osteosarcomas often pose difficulty in diagnosis to clinicians because of the overlap with other benign and malignant bone lesions. High grade osteosarcomas pose a challenge to clinician because of their propensity for recurrence. An aggressive surgical approach with post-surgical radiochemotherapy can be an effective tool in combating this variant of osteosarcoma. Repeated surgical intervention if required should be done as most patients are in the young age group. The several histopathological variants should also be recognized for proper treatment plan.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Babu H, Sharma R, Sharma VK, Aggarwal T, Rana AK. A case of recurrent chondroblastic osteosarcoma mandible. Int J Otorhinolaryngol Head Neck Surg 2021;7:698-700.