Osteosarcoma of Mandible: A Rare Report with Literature Review

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ABSTRACT
Osteosarcoma (OS) is a commonly encountered bone malignancy. It is an aggressive lesion which usually follows a fatal course. Osteosarcoma of jaw bones are rare, representing only 4 to 8% of all osteosarcomas which makes this case interesting to report. A case of OS affecting the mandible of a 35-year-old male which is rare is presented here along with literature review.

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INTRODUCTION
Osteosarcoma (OS) is a primary malignant neoplasm affecting the bone. Histologically, it is characterized by osteoblastic differentiation with the formation of tumor osteoid.\(^1\) It most commonly involves the long bones near the metaphyseal growth plates. The most common sites are femur (42%), tibia (19%), humerus (10%), jaws and pelvis (8%).\(^2\) It accounts for 15 to 36% of all primary bone tumors. It is the second most common bone malignancy after multiple myeloma.\(^3\)

Osteosarcoma of jaw bones are uncommon; since they represent only 4 to 8% of all OS.\(^4\) The posterior ramus is a common site for mandibular tumors, while the alveolar ridge, palate and sinus areas are commonly affected in the maxilla.\(^5\)

We present a rare case of OS affecting the mandible of a 35-year-old male with literature review.

CASE REPORT
A 35-year-old male reported to a private dental clinic with a chief complaint of a painful swelling in the lower right back jaw region and difficulty in mastication since 1 month. His medical history was noncontributory.

On clinical examination, a diffuse swelling was noticed at the right angle of the mandible extending below the inferior border. It extended anteroposteriorly from the premolar region up to the ramus of the mandible (Fig. 1A). The extraoral swelling was hard and tender on palpation. No lymph nodes were palpable.

Intraoral examination revealed an exophytic, ulcerated mass with distinct borders involving the alveolus in the right molar region (Fig. 1B). The mass was firm to palpate with no discharge. It was fixed to the underlying tissues.

Figs 1A and B: (A) Diffuse swelling at the right angle of the mandible and (B) intraoral view showing ulcerated swelling involving the alveolus in the right molar region
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Orthopantomogram revealed a mixed lesion with indistinct borders, root resorption of the involved teeth and perforation of the inferior border of mandible (Fig. 2). A provisional diagnosis of OS was arrived at and a differential diagnosis of aggressive ossifying fibroma was made.

On histopathological evaluation proliferating spindle shaped, plump, malignant osteoblasts (Fig. 3A) and areas of tumor osteoid were seen. High power view showed malignant osteoblasts which were pleomorphic with hyperchromatic nuclei and few areas of tumor osteoid (Fig. 3B).

A final diagnosis of osteoblastic OS was concluded.

A hemimandibulectomy was performed with no postoperative complications. The patient was discharged 1 week after the operation in good condition and was referred for radiation therapy. No recurrence has been observed yet after 6 years of follow-up.

DISCUSSION

Osteosarcomas are a highly malignant aggressive bone tumor infrequently involving the head and neck region. Maxillofacial OS occur one or two decades longer than the OS of long bones with a slight male predominance.

Osteosarcomas may arise de novo or due to any predisposing factors. The major risk factors for the development of OS of the jaws are similar to those for OS of the long bones, i.e. previous irradiation of the head and neck region, history of Paget’s disease and fibrous dysplasia. Other bone abnormalities like as chronic osteomyelitis, multiple osteochondromatosis, myositis ossificans and trauma have also been proposed as risk factors. Our patient had none of the above mentioned conditions, in our case, it could have occurred de novo or genetic factors could be suspected.

Osteosarcomas usually present as large aggressive lesions which cause considerable amount of pain and discomfort to the patient. They grow rapidly causing expansion of the cortical plates. Displacement and resorption of roots are common. They easily invade adjacent structures due to their invasive growth pattern with mucosal ulceration and pathologic fracture is common. Sensory abnormalities are encountered when the peripheral nerves are involved.

Our case presented as an aggressive lesion with perforation of the inferior border of the mandible, neurologic manifestations were not seen.

Aggressive ossifying fibroma and osteoblastoma must be considered in the clinical differential diagnosis which can resemble OS in clinical presentation and aggressive biologic behavior. Recording an accurate clinical history is important in such cases, radiography as an adjunct can help in differentiating when histopathologic features are not clear cut.

Radiographically, it can present as a mixed sclerotic lesion to give a cumulus cloud appearance or as a radiolucent lesion. In more than 50% of cases, a classic sunburst appearance is seen which is best demonstrated in computed tomography and occlusal radiographs.

This is due to osteophytic bone formation. Indistinct margins without sclerosis are a common presentation; these features were consistent in our case with a cumulus cloud appearance.
Histologically, OS can be classified according to the type of cellular component as osteoblastic, chondroblastic and fibroblastic variants. In osteoblastic type, atypical malignant osteoblasts are seen with tumor osteoid formation.

The chondroblastic type, consists of atypical pleomorphic binucleated cells composed of chondroid areas and the fibroblastic type shows atypical hyperchromatic, spindle shaped cells. Based on these features, our case was diagnosed an osteoblastic OS.

Histopathological differential diagnosis from trabecular aggressive ossifying fibroma can sometimes be a dilemma due to pleomorphism of cells and formation of osteoid which can resemble tumor osteoid of OS. The degree of pleomorphism and presence of mitotic figures are higher in osteosarcoma. Aggressive ossifying fibroma can often be differentiated from OS on the basis of radiographic appearance. The radiographic features of OS are orthoradial striations, destruction of cortices with an outgrowth of the soft tissue component, generalized widening of the periodontal ligament spaces and destruction of the lamina dura, all of which are absent in aggressive ossifying fibroma.

This differentiation is of paramount importance since aggressive ossifying fibroma is a benign lesion.

Complete surgical excision is the mainstay of treatment. Total maxillectomy/mandibulectomy or a hemimaxillectomy or hemimandibulectomy is done based on the extent of the lesion in the jaws.

Adjunct radiotherapy and chemotherapy seem to have little effect except in delaying the recurrence and does not seem to improve the survival rates significantly.

The surgical margins in OS are of prognostic importance. Patients show a better survival with surgical margins greater than 5 mm, fewer local recurrences and less metastatic disease compared to those with margins of less than 5 mm. Although achieving such a wide rim of normal tissue is a challenge in the jaws, clear margins play a role in eradication of disease and limitation of intramedullary extension.

CONCLUSION

The prognosis of OS in the jaws is poor. Our patient underwent hemimandibulectomy followed by radiotherapy, no recurrence has been observed yet for the past 6 years which makes this case interesting to report. Differential diagnosis from trabecular aggressive ossifying fibroma can sometimes be a clinical and histopathological diagnostic dilemma. Distinguishing both is mandatory since both lesions have varied prognosis.

REFERENCES