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CASE REPORT

HIDDEN WITHIN THE JAW: AN UNUSUAL FINDING OF OSTEOSARCOMA FOLLOWING MANDIBULAR TRAUMA

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ABSTRACT

This case report focuses on the aggressive nature of osteosarcoma in head and neck region which poses a diagnostic challenge and effort of maxillofacial surgeons to provide better prognosis by identifying in early stages.

Background:A 24-year-old male patient presented with post traumatic swelling in left mandible with aggressive growth in just few weeks. He was initially reluctant to undergo the procedure as he overlooked the severity of the tumour.

Diagnosis: Patient was diagnosed with parosteal juxtacortical osteosarcoma of the left mandible on clinicoradiohistopathological diagnosis and had no distant metastasis.

Treatment: Multi-modality treatment implemented with hemi-mandibulectomy tumour free margins followed by radiochemotherapy.

Outcome: There was no evidence of local recurrence and distant metastasis after one year of follow up period.

Conclusion: Osteosarcomas of head and neck region are less than 2-10% of all cases. Emphasis on early diagnosis, negative surgical margins and inclusion of radio and chemotherapy provides disease free survival.

Key words: Osteosarcoma, Mandible, Juxtacortical, Parosteal

INTRODUCTION

Osteosarcomas are rare malignant connective tissue tumours. Out of the all-primary tumours, osteosarcomas account for 20% which are mostly confirmed by the biopsy[1] 4-6 % occurs in jaw bones.[2] Osteosarcomas are of two types, primary and secondary. Etiology for the primary tumour is not known but genetic and environmental factors have a great influence. Secondary type is seen in Paget's disease, Fibrous dysplasia, craniofacial irradiation cases, Li-Fraumeni syndrome and Rottmund-Thomson syndrome.

As these tumours occur in metaphyseal growth plate of long bones, rapid bone growth acts as a major etiological factor. Other factors include genetic mutation of P53 tumour suppressor gene and mutated retinoblastoma.[1]

It occurs in 30-40 years of age groups with more of male predilection. Treatment protocols include combination of surgery, radiotherapy and chemotherapy and the 5-year survival rate for these patients is 50 to 70%.[3]

We report a case of a 24-year-old male diagnosed with osteosarcoma of left mandible accidentally found after a road accident.

Case report

A 24-year-old male patient reported with a complaint of swelling in the lower jaw on the left side of the face for one month. Swelling is not associated with pain. Patient gave a history of road traffic accident and had injury on the left side of lower jaw one month back. Patient had no difficulty related to trauma but developed a small swelling next day of trauma which grew to the size of 6x4 cm (figure 1).

Figure 1. Extraoral photograph showing swelling at the left body of the mandible

Patient reported to Department of oral and maxillofacial surgery a week after the trauma due to the swelling. On examination, general physical examination was within normal limits.

Vitals are normal. Examination of head and neck revealed facial asymmetry on left side of the lower jaw. There is a solitary oval shaped swelling of size 6 x 4 x 3 cm occupying the body of left mandible extending intraorally from 31 to 38 teeth, medially abutting the floor of the mouth and laterally gingivobuccal sulcus, inferior 3cm crossing the lower border of the mandible with no involvement of overlying skin and mucosa, tongue and floor of the mouth (figure 2). Clinically palpable supple nodes level IA and IB. Otherwise rest of the oral cavity and oro-pharyngeal examination is normal. No paresthesia or anesthesia of lower lip and chin noted. Lower lip deviated due to swelling. Margins of the swelling are distinct. Surface is slightly lobulated. No ulcerations seen extra orally or intraorally. There is no mobility or displacement of the teeth. No pus discharge. No inflammatory signs over the swelling. There is no lingual cortical expansion seen.

Figure 2. Intraoral swelling extending from 31 to 38 teeth

Panoramic radiograph, mandible showed mixed radiolucent and radiopaque pattern in the lower left quadrant in the region of 34, 35, 36 teeth. Widening of the periodontal ligament space and attenuation of the lamina dura is seen in above mentioned teeth which is the most common radiographic finding in osteosarcoma. Occlusal x-ray of mandible showed expansion of buccal cortical plate and bony spicule radiating perpendicular from the cortex of alveolar bone (figure 3).

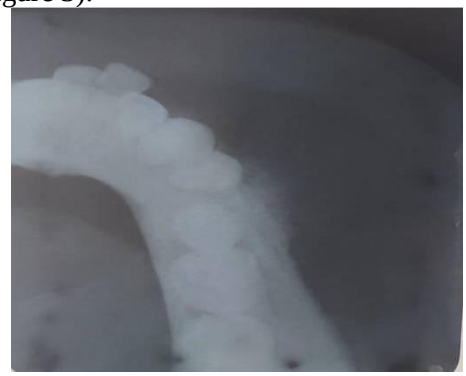

Figure 3. Occlusal radiograph of the lesion present in the left body of the mandible

Computed tomography (CT) findings suggestive of well-defined hypodense juxta cortical lesion seen encircling body of left hemimandible with irregular periosteal reaction and perpendicular bony spicules radiating from cortex, sun ray appearance suggesting parosteal osteosarcoma (figure 4).

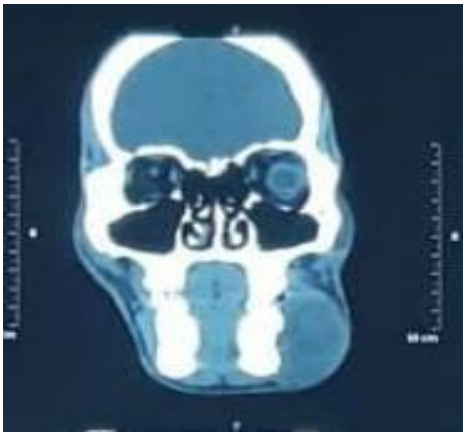

Figure 4. Coronal section of computed tomographic scan showing swelling in the left body mandible

Incisional biopsy confirmed parosteal type of osteosarcoma. Positron emission tomography (PET) scan was suggested for staging. It showed metabolically active lesion in the left half of mandible, mildly active cervical lymphadenopathy. No active regional or distant metastasis. Based on the above clinical, radiological and histopathological findings, the tumour was classified under TNM classification as T2N0M0, we maxillofacial surgeons with the help of the oncologists, left hemi-mandibulectomy was planned with normal margins of 1.5 cm along with left modified neck dissection with preservation of internal jugular vein and spinal accessory nerve (figure 5,6) and reconstruction with pectoralis major myocutaneous pedicled flap for intraoral defect coverage.

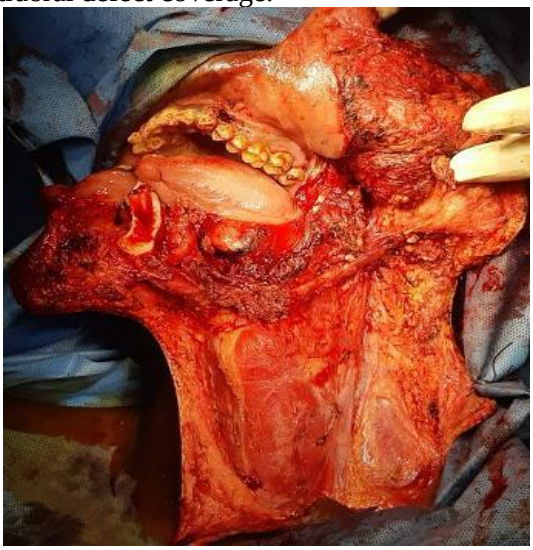

Figure 5. Left Composite resection hemi-mandibulectomy and modified neck dissection

Post-operative period was uneventful. Specimen was sent for histopathological examination to see surgical margins are free of tumour remnants. Excisional biopsy also revealed parosteal osteosarcoma T2N0M0.

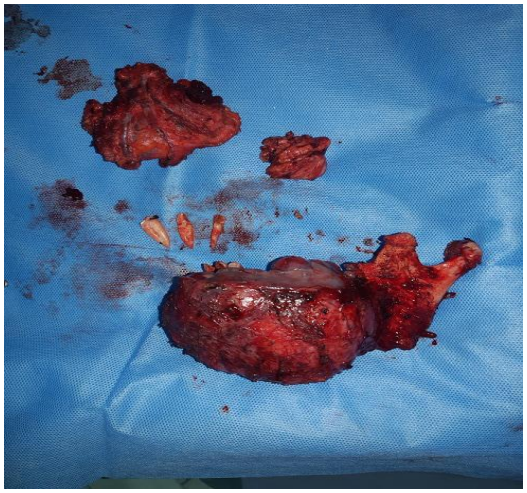

Figure 6. Specimen after composite hemi-mandibulectomy and modified neck dissection

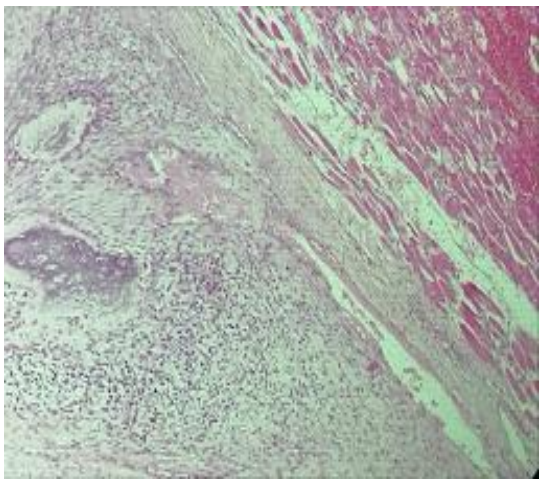

Figure 7. Histopathological section of the specimen

Radiotherapy was given for 6 weeks, 60 gray each session. Adjuvant chemotherapy with 5 cycles of CISPLATIN 120mg/m2 IV D1, ADRIAMYCIN 100mg/m2 IV D1 for improved prognosis(figure 8, 9)..

One year follow up period patient showed no signs of recurrence.

There is a plan to take up for second stage surgery for reconstruction with fibula bone graft.

Figure 8. Patient images for Adjuvant therapy

Figure 9. Intraoral images for Adjuvant therapy

DISCUSSION

This case is very peculiar and surprising that he reported to the Department of oral and maxillofacial surgery with history of road traffic accident and injury to the left side mandible and his orthopantomogram revealed a highly malignant and rare tumour osteosarcoma hidden inside the jaw. He presented with a small swelling with no symptoms at the left body of the mandible. There was rapid growth pattern of the lesion day after day and increased to the size after incisional biopsy.There was no paresthesia of the lower lip in this case, as the involvement of inferior alveolar canal indicates poor prognosis. Studies show that there is microscopic spread seen through mandibular canal, mental nerve and periodontal ligament. Inferior alveolar canal facilitates rapid spread to the adjacent soft tissues.[10] The orthopantomogram, occlusal and CT scan revealed characteristic feature of stippled bone pattern and destruction of the cortical plate and perpendicular bony spicules radiating from the cortex. Characteristic sun ray appearance was seen in this case. CT provides excellent detection of tumour calcification, cortical involvement, soft-tissue and intramedullary extension.[6] MRI is even more effective in demonstrating the intramedullary and soft tissue components. There are two clinical variants i.e., Juxtacortical and Intramedullary. Juxtacortical has parosteal and periosteal types. Histologically based on amount of osteoid cartilage or collagen fibers which are produced by the tumour they are osteoblastic, fibroblastic and chondroblastic.[4,5,7] Incisional biopsy revealed as parosteal chondroblastic variant of osteosarcoma. Chest X-ray and PET-CT of this case revealed no metastasis to the lung or other regions. Metastasis is lower in jaw bones compared to long bones. Metastasis is higher in post irradiation osteosarcoma lesions of jaws.[9] Biological behavior of jaw lesions is different from long bones even though they share similar histological features. For the tumours of maxilla, maxillectomy is performed but difficulties arise due to involvement of structures like maxillary sinus, pterygopalatine fossa and orbital floor. Obturators are used to cover the large defects which are categorized into surgical, post-surgical and definitive.[3]

There are special histological investigations such as bone matrix protein, osteocalcin bone protein which are useful in diagnosing osteosarcomas from other malignant tumours. Anatomical limitations in maxillofacial region cause difficulties in acquiring tumour free margins leading to local recurrence of 33-39% for which multimodality therapy of radical resection with 1.5- 2 cm normal margins followed by radiotherapy and chemotherapy which effects the better prognosis of the lesion.[6,8] Presurgical chemotherapy or radiotherapy is preferred in large tumours to reduce the volume and less aggressive surgical treatment.[8] Radiotherapy is opted for cases with residual, un-ressectable and recurring lesions.[2]

CONCLUSION

This case enlightens us that sudden bony growth in the jaws should alert and be viewed with suspicion of malignant lesion like sarcomas and treated radically with combination therapy for better prognosis and improved survival rate.

DECLARATION

Ethics approval and consent to participate
Not applicable
Patient consent for publication
Not applicable
Animal studies
Not applicable
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Conflicts of interest and financial disclosures

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