IOSR Journal of Dental and Medical Sciences (IOSR-JDMS)
e-ISSN: 2279-0853, p-ISSN: 2279-0861.Volume 17, Issue 3 Ver.9 March. (2018), PP 34-41
www.iosrjournals.org

Osteosarcoma of the Mandible: Case Report And Review Of Literature

Dr. Pushan Maity[1], Dr. Amit Ray[2], Dr. Monika Gupta[1]
[1](PG Student, Department of Oral and Maxillofacial Surgery, Guru Nanak Institute of Dental Sciences and Research, Kolkata, India)
[2](Professor and Head, Department of Oral and Maxillofacial Surgery, Guru Nanak Institute of Dental Sciences and Research, Kolkata, India)
Corresponding Author: Dr. Pushan Maity

***Abstract**: Osteosarcomas are malignant neoplasms arising from mesenchymal stem cells or their early progenitor cells.The incidence of osteosarcoma is low in malignant tumors; however it is the most common primary tumor of bone.Conventional osteosarcoma has been classified depending on the predominant type of extracellular matrix into subtypes - osteoblastic, chondroblastic, fibroblastic.Osteosarcomas of the jaws have a similar male and female pedilection. Mandibular lesions are more frequent than those in the maxilla. The radiographic appearance of Osteosarcoma depends on the proportions between bone destruction, calcification, new bone production and periosteal new bone formation. Tissue biopsy is needed for a definitive diagnosis. The specimen should be taken from the center of the lesion to avoid including reactive periosteal bone in the specimen.Surgery is the primary treatment for Osteosarcoma of jaws but it cannot be depended as the sole treatment modality.Chemotherapy (CT) has become an important therapeutic adjuvant in the treatment of osteosarcomas of all sites. Here we present a case of Fibroblastic Osteosarcoma of the Mandible with a review of Literature.*

Date of Submission: 03-03-2018 Date of acceptance: 19-03-2018

I. Introduction

The incidence of osteosarcoma is low in malignant tumors; however it is the most common primary tumor of bone[1]. Osteosarcoma of jaw differs from Osteosarcoma of the long bones in its prognosis, presenting a lower incidence of metastasis, as well as better survival.[2,3,4]

Here, we present a case report of Osteosarcoma in the mandibular region of a 50 years old Male patient along with review of literature.

II. Case Report

A 50 years old Male(62kg) reported to the Department of Oral and Maxillofacial Surgery, GNIDSR, Kolkata with complains of a swelling in the lower front teeth region for the last 1 year on April, 2017.

On taking the history it was found that the patient had a trauma when some heavy object fell on his face approximately 35 years ago. On radiographic examination, his then physicians found a fracture of the right body of mandible. However the due to the patient's reluctance, no definitive treatment was done. According to the patient the area of trauma healed uneventfully without any symptoms. After that about a year ago, he noticed pain in the lower front teeth and within a few weeks he noticed mobility of those teeth. The patient took some Over the Counter analgesics as well as prescription medication advised by his physician which provided brief episode of pain relief but failed to impart any long term solution. After a month or so he noticed a soft swelling near the lower front teeth which slowly grew in size subsequently. The area was not painful and no incidence of bleeding or pus discharge occurred. Any episode of accelerated growth or spontaneous remission was also absent.

The patient was of average build and normal gait. All vital signs were normal. There was no abnormal breath sounds and the chest x-ray appeared normal. No cervical, axillary or inguinal lymphadenopathy was present.

On extraoral local examination, a swelling was present on the base of the lower lip near the Labiomental crease, on the midline and the overlying skin appeared normal with no discolourations or ulceration. There was no presence of any discharging sinus. The Swelling was soft , non pulsatile, non tender, fixed to the underlying structure, overlying skin was free, no local rise of temperature was noted. Mouth opening was normal(45mm), no deviation of mandible was present on movement. Facial nerve functions were normal. No lip paraesthesia was present.

The Lower border of mandible was expanded, with hard lobulated surface from left to right angle of the mandible. No clicking sound on bilateral TMJ auscultation was heard. No bruit on surface of the swelling was present.

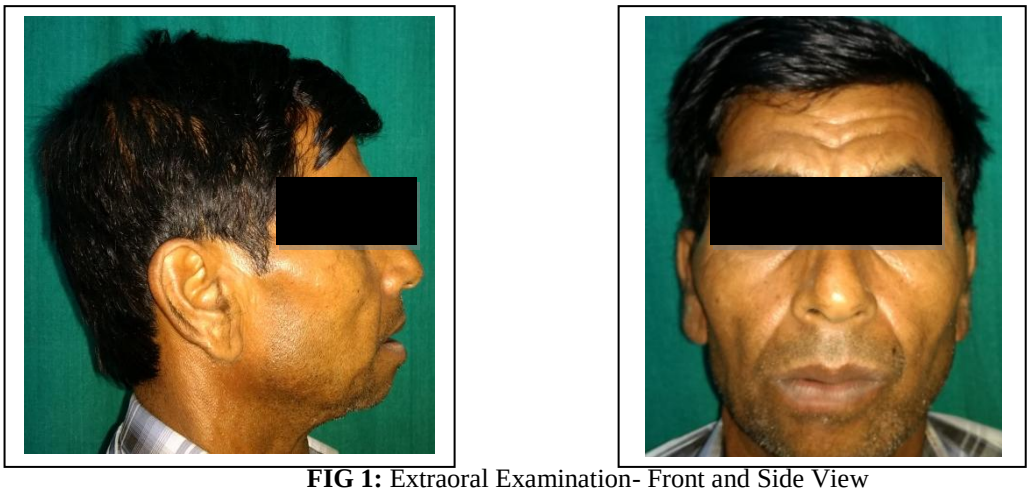

FIG 1: Extraoral Examination- Front and Side View

On intraoral examination, a localized sessile swelling was present in the lower incisor region in the midline on both buccal and lingual aspect extending from the right lower canine to the left lower canine. The overlying mucosa appears normal except 2 lobules present on the surface. The Swelling on the midline was soft, non pulsatile, non tender, mobility of lower incisor teeth were present. The buccal swelling involved the lower gingivobuccal sulcus. The Floor of the mouth was not involved by the lingual side of the lesion. Another Diffuse swelling was present near the buccal gingiva in relation to the right lower molar teeth. The Distal swelling was firm and lobulated. No pus discharge was noted. The tongue movements were normal. Hard, Lobulated surface was throughout the mandible body in both buccal and lingual aspect.

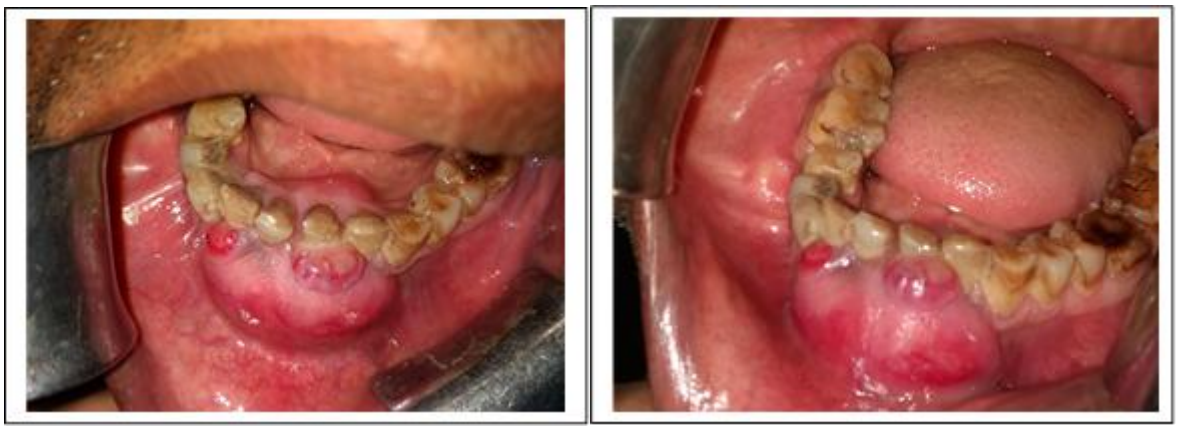

FIG 2: Intraoral examination showing the swelling on the midline as well as the Right Molar region.

On Radiological examination, Orthopantomogram showed diffuse area of mixed radiolucency along the body of mandible on both sides, with irregular margin devoid of sclerosis. Destruction of lower border of mandible was seen on the right mandibular body region. No root resorption of teeth was seen. However, there was some displacement of teeth in the left lower Canine-Premolar region resulting in an increased interdental space between the left lower first molar and second premolar. Widening of the Periodontal spaces (Garrington sign) was absent. Notably, irregular trabecular bone patter was seen throughout the mandible as well as the maxilla.

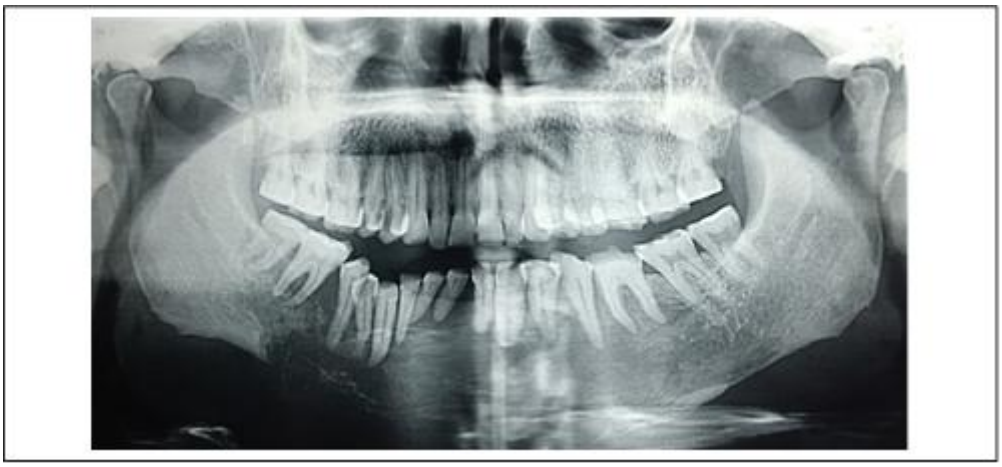

FIG 3: Orthopantomogram

Contrast enhanced CT scan shows a well demarcated homogenous swelling on the midline, without any ossification, areas of enhancement noted with enhancing border. Mixed radiolucency was seen throughout the body of the mandible extending to the lower border. There was expansion and thinning of buccal and lingua cortices with perforation of the buccal cortex in the midline. Irregular trabecular bone pattern was present on the mandible as well as the maxilla. Interestingly, both the maxillary sinuses were obliterated by cancellous bone with irregular trabecular pattern. No lymphadenopathy was seen.

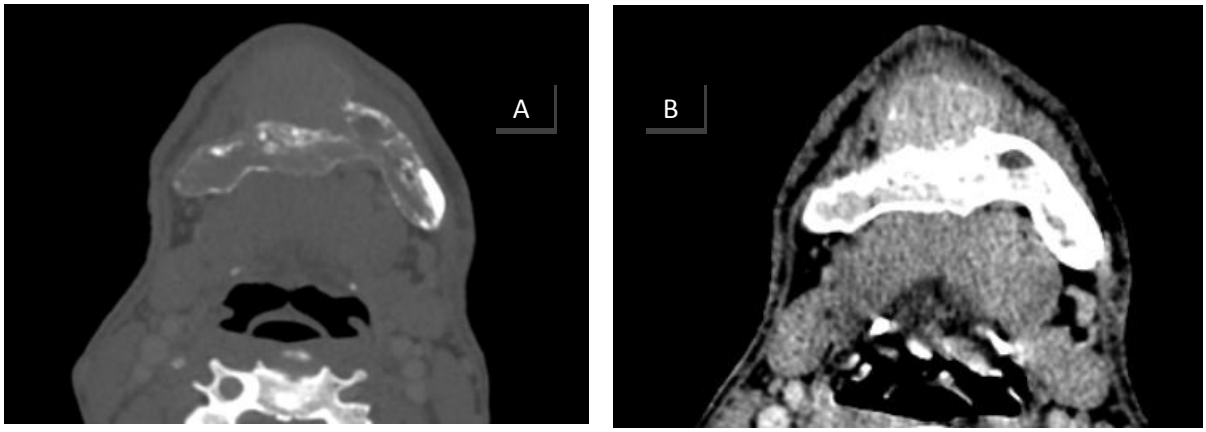

FIG 4: Contrast Enhanced CT scan in A) bone window, B) soft tissue window

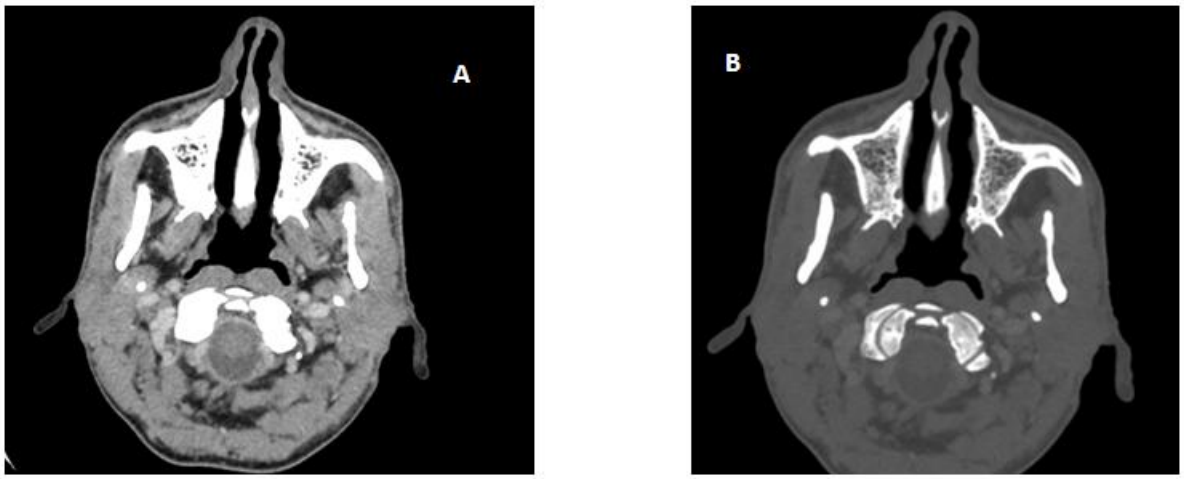

FIG 5: Contrast Enhanced CT scan showing obliterated maxillary sinus in A) soft tissue window, B) bone window

An incisional biopsy was taken from the midline as well as a cortico-cancellous bone specimen from both the right and left body of the mandible. The Histopathological examination revealed presence of Osteosarcoma- fibroblastic variant in the specimen taken from the midline and right body of mandible. The specimen from the left body of mandible showed reactive changes.

Treatment:

Submandibular and intraoral incision was placed on the buccal as well as the lingual sulcus. Segmental mandibulectomy was done from left angle to the right angle. Reconstruction of the floor of the mouth was done using Submental flap. Elective tracheostomy was done and a 7.5mm cuffed tracheostomy tube was placed.

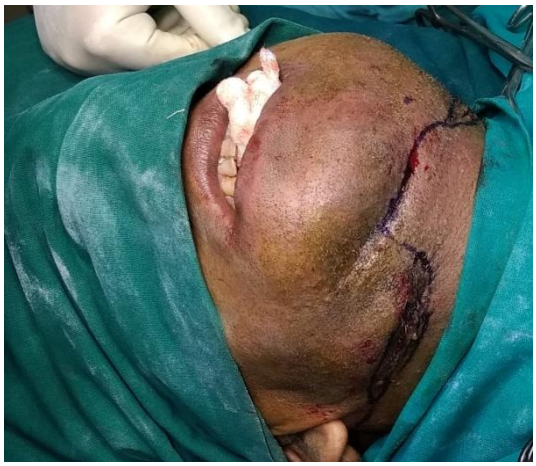

FIG : 6. Incision Marking

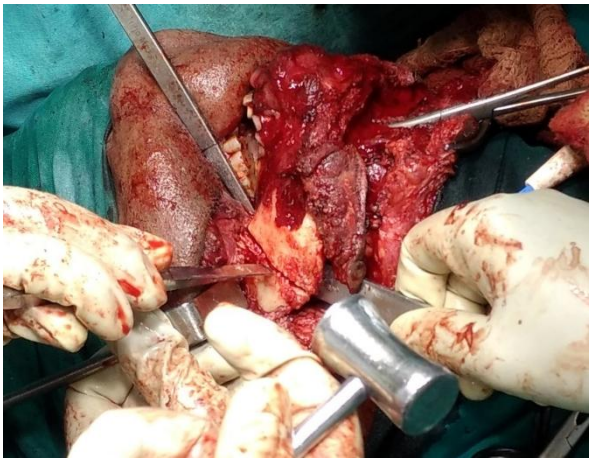

FIG 7. Osteotomy Cut for Resection

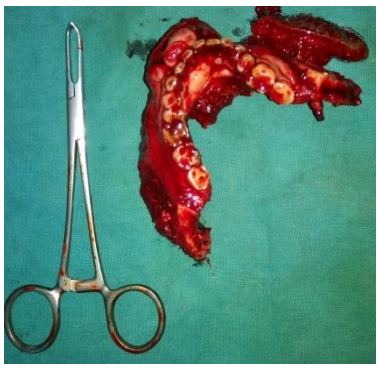

FIG 8: Surgical Specimen

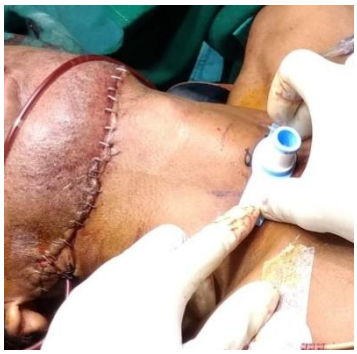

FIG 9. Tracheostomy Tube in place

Postoperative Histopathology showed marked fibroblastic proliferation. Few Giant cells were seen close to tumour osteoid. The diagnosis of Osteosarcoma – Fibroblastic variant was made.

Postoperative period was uneventful with regular irrigation, dressing and tracheostomy care. The tracheostomy dependence was weaned off sequentially and de-cannulation was done by the end of one week postoperatively. No hindrance of tongue movement persisted. The patient was referred for subsequent chemoradiotherapy.

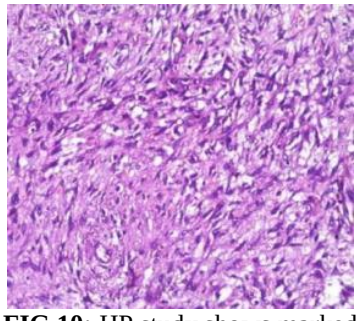

FIG 10: HP study shows marked fibroblastic Proliferation

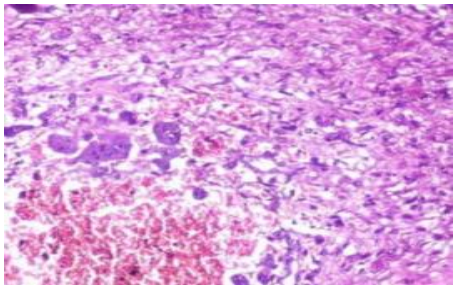

FIG 11. HP study showing few Giant Cells

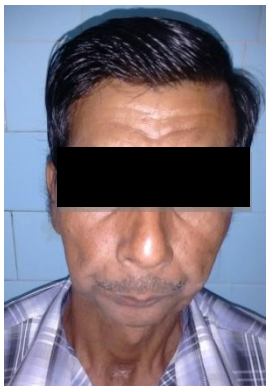

FIG 12: Post-Operative Appearance of the Patient

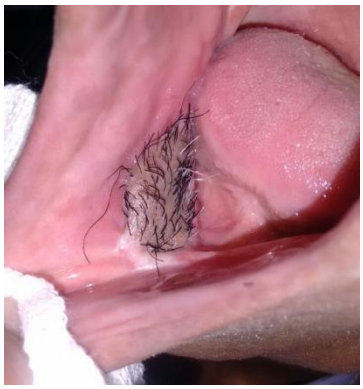

FIG 13. Healed Submental Flap showing Hair growth

III. Discussion

Osteosarcomas are malignant neoplasms arising from mesenchymal stem cells or their early progenitor cells. A definitive feature of Osteosarcoma is the ability of the malignant cellular stroma to produce malignant osteoid, which is unique other messenchymal tumors.[4]

Klein et al. classified Osteosarcoma as[1]- Osteosarcoma Types

- Central
  - High-grade
    - Conventional
    - Telangiectatic
    - Small cell
    - Epithelioid
    - Osteoblastoma-like
    - Chondroblastoma-like
    - Fibrohistiocytic
    - Giant cell–rich
  - Low-grade
    - Low-grade central
      - Fibrous dysplasia–like
      - Desmoplastic fibroma–like
- Surface
  - Low-grade
    - Parosteal
  - Intermediate-grade
    - Periosteal
  - High-grade
    - Dedifferentiated parosteal
    - High-grade surface
- Intracortical
- Gnathic
- Extraskeletal
  - High-grade
  - Low-grade

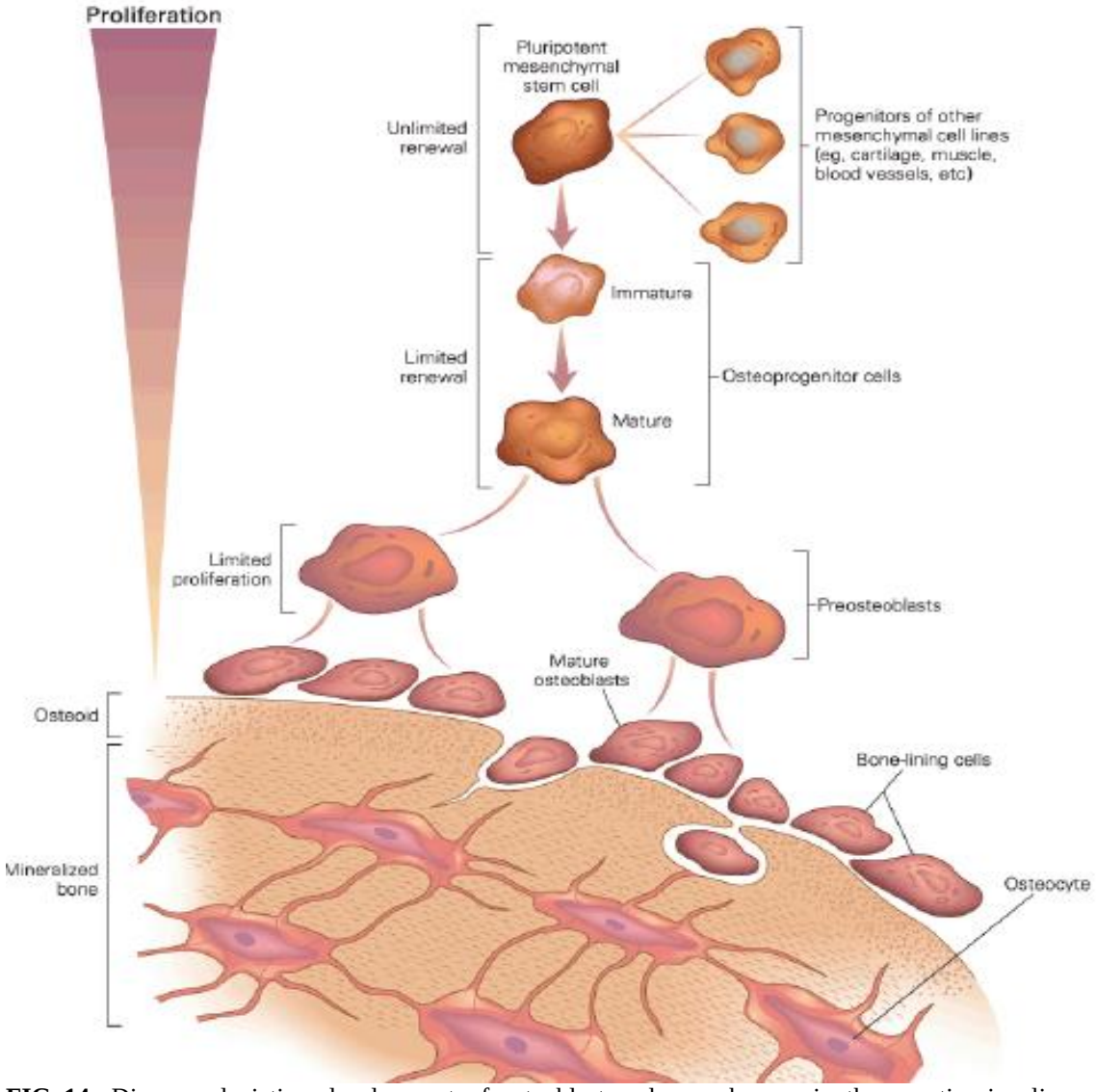

FIG 14: Diagram depicting development of osteoblast and any changes in the genetic signaling pathway controlling this process can lead to development of Osteosarcoma[4]

Conventional Osteosarcomas are most easy to diagnose. Histopathology shows spindled or polyhedral tumor cells; pleomorphic and hyperchromatic nuclei. The produced extracellular matrix by the tumor cells may be osseous, cartilaginous, or fibrous. Conventional osteosarcoma has been classified depending on the predominant type of extracellular matrix into subtypes - osteoblastic, chondroblastic, fibroblastic.[1]

Osteosarcomas of the jaws have a better prognosis than osteosarcomas of long bones. Commonly treatment failure results from local recurrences followed by lung metastasis, brain metastasis, and then metastasis to other bones. Distant metastases occurs via Hematogenous route to the right heart, subsequently pumped into the capillaries of the lungs, or retrograde flow to the brain or other bones.[4] Osteosarcomas of the jaws have a similar male and female pedilection. Mandibular lesions are more frequent than those in the maxilla. Parosteal or juxtacortical osteosarcomas arise from periosteum.[4] Surface osteosarcomas occur at a later age as than conventional osteosarcomas. The common site is the lower end of the femur followed by the upper end of the tibia and upper end of humerus, respectively. The periosteal variant affects the tibia more commonly than the parosteal variety. Neo-adjuvant chemotherapy is the standard of care for high grade surface osteosarcomas. Parosteal osteosarcomas are low grade lesions and can be treated by surgery without chemotherapy.[5]

The clinical feature of Osteosarcoma of long bones is pain during movement. However, in jaw lesions pain is not a prominent feature and instead swelling is commonly the presenting complaint. Most patients notice the tumor after dental treatments , mainly extractions. This likely due to the rapid growth of tumor immediately after tooth extraction.[6]

The radiographic appearance of Osteosarcoma depends on the proportions between bone destruction, calcification, new bone production and periosteal new bone formation[7]. Lesions may be osteolytic, osteogenic, or mixed. If the tumor invades the periosteum, new bone may develop perpendicular to the surface of the lesion producing sun ray appearance. The widening of periodontal ligament space and inferior dental canal, together with sunburst effect are almost pathognomonic of Jaw Osteosarcoma.[2] Contrast Enhanced Computerized tomography (CT) and magnetic resonance imaging can be used to assess the size of the lesion for staging extramedullary involvement, calcification and invasion into adjacent tissues such as pterygopalatine fossa, infra temporal fossa and cranial cavity.[5]

Tissue biopsy is needed for a definitive diagnosis. The specimen should be taken from the center of the lesion to avoid including reactive periosteal bone in the specimen, which could lead to a misdiagnosis.[4] Histopathologically, Ostreosarcoma of long bones and jaw bones share common features. The diagnosis is based on the recognition of osteoid production by tumor cells. There may be chondroblastic or fibroblastic elements. Depending on the predominant type of extracellular matrix present, Osteosarcoma are categorized histopathologically into osteoblastic, chondroblastic, or fibroblastic subtypes.[8]

The differential diagnosis of osteosarcoma from other sarcomas by Immunohistochemical studies (eg, malignant fibrous histiocytoma, fibrosarcoma, Ewing's sarcoma) is important because of the specific therapy available for osteosarcoma patients. Most osteosarcomas express vimentin and some tumors focally express cytokeratin and desmin. Bone matrix proteins, such as osteocalcin, alkaline phosphatase, and osteonectin, are expressed in osteosarcomas. However, their presence has also been detected in chondrosarcomas, Ewing's sarcoma, fibrosarcomas, and malignant fibrous histiocytomas. Caution should also be used in the interpretation of focal expression of a variety of markers (eg, S-100, actin, epithelial membrane antigen) found occasionally in otherwise typical osteosarcomas. Extraskeletal osteosarcomas of the fibroblastic subtype often have sparse amounts of osteoid and can be differentiated from malignant fibrous histiocytoma on the basis of strong expression of alkaline phosphatase. Chondroblastic osteosarcoma and chondrosarcoma, however, cannot be distinguished immunohistochemically. Furthermore, it remains to be seen if the expression of CD31 or CD34 helps in the differential diagnosis between telangiectatic osteosarcoma and angiosarcoma. The different types of collagen present in the bone matrix are also produced by other tumors and therefore have no application in differential diagnosis. However, recent reports suggest that the basic calponin gene, a smooth muscle differentiation-specific gene that encodes an actin-binding protein involved in the regulation of smooth muscle contractility, is expressed in osteosarcomas and that this expression may have favorable prognostic implications.[9]

Surgery is the primary treatment for Osteosarcoma of jaws but it cannot be depended as the sole treatment modality[1,2]. Survival is favorable when surgical margins are negative. Surgery involves resection of the entire tumor with wide margins of at least 3 cm from the clinicoradiographic edge or to the joint or the nearest suture if in the midface. A neck dissection is not required because, as is typical of most sarcomas, osteosarcomas do not metastasize via lymphatics except in rare instances.[4] According to August et al, clear surgical margins are statistically associated with survival.[10] Maxillary lesions are often difficult to be treated as involvement of maxillary sinus, pterygopalatine fossa and orbital fossa often hides the spread of tumor until extensive spread. Often, maxillectomy is required.[5] If cervical lymphatics are involved, neck dissection is needed[2]. Chemotherapy (CT) has become an important therapeutic adjuvant in the treatment of osteosarcomas of all sites[5]. Rosen et al reported 93% recurrence-free survival at a median follow-up of 20 months, in long bones, using preoperative and postoperative chemotherapy with surgery. The commonly used chemotherapeutic agents are doxorubicin, cisplatin, adriamycin and methotrexate. Chemotherapy was effective in treating subclinical metastases in Osteosarcoma of long bones and the 5 year survival for patients treated with surgery alone was 15%, but 60–80% for patients treated with surgery and chemotherapy.[11] The most used procedure is Rosen's protocol/ Sandwich technique, which includes preoperative Chemotherapy, surgery, and postoperative Chemotherapy[5,11]. The excisional biopsy determines the accuracy of excision and the response of tumor to preoperative CT, in comparison to the incisional biopsy. This assists in the selection of the postoperative chemotherapeutic treatment.[5]

Because the concern is recurrence at the primary tumor site and metastasis to the lungs, a clinical examination and a chest radiograph are recommended every 4 months for the first 2 years and then every 6 months for the next 3 years. A PET scan should be used in preference to a chest radiograph if available. Such examinations should be carried out on an annual basis. Any suspicious signs seen on a plain chest radiograph or a PET scan should be evaluated with a HRCT of the chest.[4]

IV. Conclusion

The exact etiology of osteosarcoma of the jaws is not known. The presence of a mass is the most common presenting symptom. Sometimes patients present with dental symptoms such as loose teeth. Paraesthesia may be a presenting symptom of mandibular osteosarcoma. Role of Immunohistochemistry is gaining importance as it can aid in selection of treatment methods as well as serve as a prognostic indicator. Surgical excision with adjuvant chemotherapy has better survival than in the case with osteosarcomas of long bones.

References

[1]. Klein MJ, Siegal GP. Osteosarcoma. American Journal of Clinical Pathology. 2006;125(4):555–81.
[2]. Garrington GE, Scofield HH, Cornyn J, Hooker SP. Osteosarcoma of the jaws.Analysis of 56 cases. Cancer. 1967;20(3):377–91.
[3]. Regezi JA, Zaro RJ, Mcclatchey KD, Courtney RM, Crissman JD. Osteosarcomas and chondrosarcomas of the jaws: Immunohistochemical correlations. Oral Surgery, Oral Medicine, Oral Pathology. 1987;64(3):302–7.
[4]. Marx RE, Stern D. Oral and maxillofacial pathology a rationale for diagnosis and treatment. Hanover Park, IL: Quintessence Pub. Co.; 2012.
[5]. Khan S, Barwar N, Kumar V. Surface osteosarcomas: Diagnosis, treatment and outcome. Indian Journal of Orthopaedics. 2014;48(3):255.
[6]. Nissanka E, Amaratunge E, Tilakaratne W. Clinicopathological analysis of osteosarcoma of jaw bones. Oral Diseases. 2007;13(1):82–7.
[7]. Unni, K. Krishnan, Carrie Y. Inwards. Dahlin's bone tumors: general aspects and data on 10,165 cases. Philadelphia: Wollters Kluwer Health/Lippincott Williams & Wilkins.; 2010.
[8]. Schajowicz F. Histological Classification of Bone Tumours. Histological Typing of Bone Tumours. 1993;:3–6.
[9]. Kim J, Ellis GL, Mounsdon TA. Usefulness of antikeratin immunoreactivity in osteosarcomas of the jaw. Oral Surgery, Oral Medicine, Oral Pathology. 1991;72(2):213–7.
[10]. August M, Magennis P, Dewitt D. Osteogenic sarcoma of the jaws: factors influencing prognosis. International Journal of Oral and Maxillofacial Surgery. 1997;26(3):198–204.
[11]. Rosen G, Caparros B, Huvos AG, Kosloff C, Nirenberg A, Cacavio A, et al. Preoperative chemotherapy for osteogenic sarcoma:Selection of postoperative adjuvant chemotherapy based on the response of the primary tumor to preoperative chemotherapy. Cancer. 1982;49(6):1221–30.

Dr. Pushan Maity "Osteosarcoma of the Mandible: Case Report And Review Of Literature."IOSR Journal of Dental and Medical Sciences (IOSR-JDMS), vol. 17, no. 3, 2018, pp 34-41.