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OSTEOSARCOMA OF MANDIBLE - A CASE REPORT AND CBCT FEATURES

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ABSTRACT

Osteosarcoma (OS) is a malignant connective tissue tumor originating from bone, which usually presents with an aggressive clinical course. It is a primary bone malignancy of long bones and shows rare occurrence in jaw bone. The aim of this case presentation is to demonstrate role of CBCT in diagnosing such highly malignant tumor of jaw bone with varied radiographic feature, even before its histopathological confirmation. This will help in early diagnosis and treatment planning.

INTRODUCTION

Osteosarcoma is malignancy of mesenchymal cells which have the ability to produce osteoid or immature bone [1]. Neville has classified OS as central (arising within the medullary cavity), surface (arising within juxtracortical region), extraskeletal (arising within soft tissue) [1]. Etiology of OS is largely unknown, but adolescent growth spurts, rapid bone growth and hormonal factors may play important role. Predisposing factors may include radiation exposure, alkylating agent, Paget disease of bone, fibrous dyaplasia, Li-Fraumeni syndrome, hereditary retinoblastoma, Rothmund Thompson syndrome, alteration in p53, RB1 and q21. The current knowledge on OS of jaw has indicated that certain other factors which can correlate with the occurrence of OS included linear bone growth and genetic and environmental factors [2].

OS accounts for 20% of all sarcomas and only 6% affects head and neck region [1, 3]. OS of jaw has a broad age range but peak occurrence is in third through fifth decade of life with slight male predilection [1]. It may affect either maxilla or mandible but has a slightly higher predilection for the mandible. Mandibular tumors develop more in the body, followed by angle, symphysis, and ramus areas where as maxillary tumors arise frequently in alveolar ridge, palate, floor of maxillary antrum, and may even involve zygoma and orbital rim [1].

Patients with OS of jaw usually report to dentists with symptoms such as swelling, pain, tooth mobility, ill-fitting dentures and paresthesia. They may often get misdiagnosed as periapical lesions, impactions or odontogenic lesions due to non-specific symptoms [4]. Therefore radiographic evaluation plays important role in diagnosis of OS. Key radiographic features of OS are widening of periodontal ligament space, spiking roots, codman's triangle, sunburst appearance [1]. Advanced imaging technology like CBCT is of great help in assessing bone destruction and osteogenesis pattern in OS. This may ease early diagnosis and treatment planning for OS patient.

Case Report

A 20-year-old female patient reported to Department of Oral Medicine and Radiology with the chief complaint of pain and swelling in the left side of the lower jaw, the duration of the complaint was of one and half months. The patient was apparently alright one and half month before when she underwent extraction of lower left second molar which was painful. The swelling started after the extraction and increased aggressively to a large size.

Extra-oral examination revealed a large diffuse swelling on lower half of the left side of the face, extending antero-posteriorly from the corner of mouth to 2cm from angle of mandible and supero-inferiorly from zygoma to1cm below the inferior border of mandible. Skin was intact throughout and no pus discharge were noted [Fig.1A,1B]. On palpation, the

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swelling was bony hard, warm and tender. It was non movable in both the antero-posterior and the lateral directions. The sub-mandibular lymph nodes were enlarged, hard and tender.

Intra-oral examination revealed a large exophytic fleshy soft tissue growth on left side, extending from retromolar pad area to the distal aspect of mandibular left canine. Baccally swelling was extending along the buccal vestibule and causing its obliteration. Lingually it was extending to floor of the mouth, causing obliteration of lingual vestibule. Surface of lesion was ulcerated and erythematous having indentations of opposing teeth and was covered with white slough. Certain areas showed hemorrhagic surface due to increased vascularity as compared to normal mucosa [fig.2]. Borders of swelling were well defined and firm in consistency. Swelling was tender on palpation. Oral hygiene of patient was poor, with the chunks of calculus were noted on teeth. Patient was unable to occlude teeth due large swelling making it difficult to chew and swallow food.

Figure1, A Clinical image showing, extra oral swelling in left mandibular region. B. Note the dryness and scaling on skin as shown by arrow.

Figure 2 Clinical picture showing, fleshy growth intraorally in left mandibular region obliterating buccal and lingual vestibule. Note the change in color of mucosa due to increased vascularity as compared to right side. Superior surface showing indentation of opposing teeth and whitish slough.

On radiographic examination, Orthopantomogram showed diffuse irregular bone loss in the region of 34- 38. Roots of these teeth showed tapered narrowing which is also termed as 'spiked' root resorption. There was widening of PDL space seen on the mesial aspect of 33, 34, 35 & 36 [fig.3]. Caldwell view showed diffuse bone loss with intact buccal cortex and the shadow of bulging soft tissue on left side [fig.4]. As radiographic picture was unusual, the patient was subjected to CBCT to study extent and nature of the lesion.

CBCT findings showed a mixed isodense and hyperdense lesion extending from left mandibular canine to the mid ramus region. Margins of lesion were ragged and infiltrating. Axial section showed tiny perforation of buccal and lingual cortical plates [fig.5].

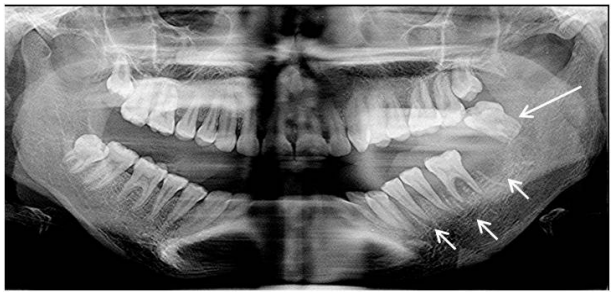

Figure 3 OPG showing, diffuse bone loss in 36, 37 & 38 region. Also note PDL widening of 34,35,36 and displacement of 38.

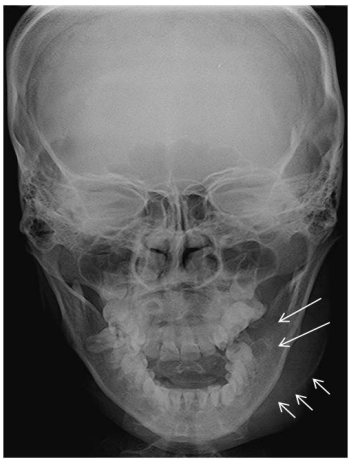

Figure 4 Caldwell view showing, diffuse bone loss with intact buccal cortex, as shown by long arrows. Note the shadow of bulging soft tissue on left side as shown by short arrows.

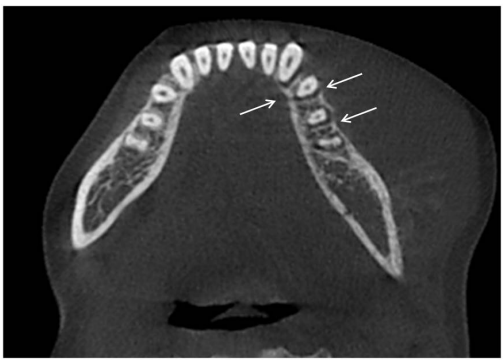

Figure 5 Axial CBCT view showing, multiple tiny perforation of buccal and lingual cortex in the area of lesion.

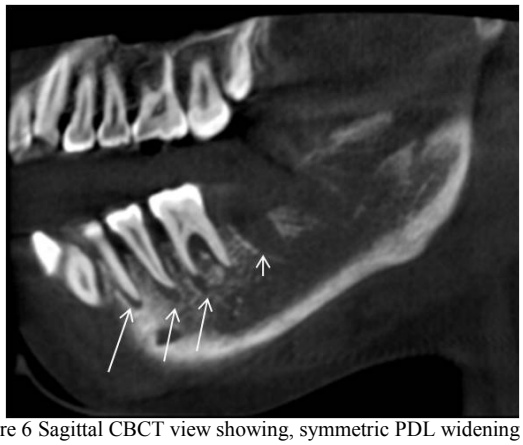

Figure 6 Sagittal CBCT view showing, symmetric PDL widening with 34, 35, 36. And extraction of 37.

The sagittal section showed very clearly highly irregular bone loss involving the left body of the mandible with islands of normal bone scattered in between. Unilateral widening of the periodontal ligament space can also be appreciated in 34, 34, 35 & 36.Extraction socket of 37 also appreciated in sagittal section [fig.6].

Coronal section showed periosteal new bone formation in the form of faint radiopaque bony striae which were radiating perpendicular to the outer cortex of alveolar bone of the mandible giving sunburst appearance to the lesion [fig.7B]. The same picture can be appreciated from axial section [fig.7A]. This finding was not recorded on Orthopantamogram and Caldwell view.

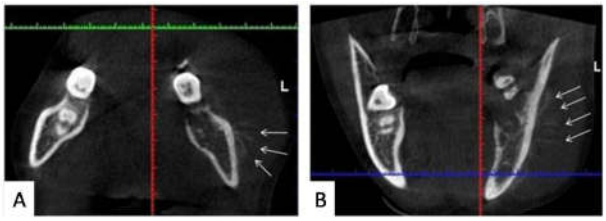

Figure7 A & B showing, Axial & coronal section of CBCT at the level of molar-ramus region showing faint bony spicules radiating perpendicular to the outer cortex giving sun burst or sun-ray appearance.

3D construction showing ample amount of bone loss on left side of mandible as compared to the normal right side and floating 38 was also noted [fig. 8A, 8B].

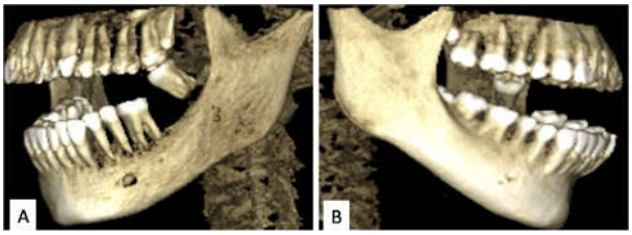

Figure 8 A & B showing 3D comparison of left and right side showing reduced bone density in left mandibular body and ramus compared to right side.

Correlating the clinical and radio- graphic features, a provisional diagnosis of osteosarcoma involving the jaw bone was made. Incisional biopsy showed florid atypical stromal proliferation, vascular proliferation, hemorrhage, chronic active nonspecific inflammation and focal cartilaginous tissue. Patient was then referred to the oncology department for further evaluation of other bones and organs for metastasis and treatment. But due to sad demise of patient before any treatment we were not able to follow up any investigation and treatment.

DISSCUSSION

OS is an aggressive malignant tumor of jaw represented with the general symptoms such as, swelling, pain and parasthesia. The radiographic differential diagnosis of OS of jaw include the lesions showing mixed radio-opaque and radiolucent appearance such as fibrous dysplasia, chronic osteomylitis, myositis ossification, cement-osseous dysplasia, osteoma [5-7]. On Conventional radiographs it is difficult to diagnose and differentiate between benign fibro-osseous lesions, inflammatory lesions and malignant tumors due to the superimposition of details of the complete anatomic region radiographed. CBCT eliminates superimposition of the structures and allows better three dimensional observation of the lesion.

Fibrous dysplasia shows Superior displacement of the mandibular canal and a fingerprint bone pattern which are pathognomonic for FD. Other features of FD are displacement of the sinus cortex, alteration of the lamina dura to the abnormal bone pattern, and narrowing of the periodontal ligament space. FD also shows enlargement of affected jaw bone due to expansion of cortical plates. Most important feature of FD is that the border of lesion blends with the normal bone, unlike OS which shows permeative borders.

Osteomyelitis shows sequestra formation which differentiates it from other lesions. Periosteal bone formation in Osteomyelitis occurs in multiple lamination form, unlike OS where it occurs as spiculations perpendicular to the periosteum.

Osteosarcoma may destroy lamina dura and show rapid progression in soft tissue like periodontal ligament which leads to the widening of the ligament space. This Symmetric widening of the periodontal ligament space is called as Garrington sign which is well appreciated on the sagittal sections of CBCT. This is an important early feature of bone malignancies like in OS, chondrosarcoma and Ewing's sarcoma [9].

Sunburst appearance is another key feature of OS of jaw which occurs due to perpendicular alignment of spicules of newly formed dysplastic osteoid to the jaw bone. It is indicative of fast biologic process and represents a disturbance in reparative phase of bone formation [5]. In present case conventional radiograph didn't show such features which were well appreciated on both the coronal and axial sections of CBCT.

Codman's triangle is a tent-like elevation of periosteum caused due to periosteal reaction at periphery of tumor. New reactionary bone is soon destroyed by pathologic process leaving only a rim of new bone supporting the periosteum, leads to the formation of Codman's triangle [10].

Bone destruction and bone formation pattern of OS can be best studied on CBCT, which give an insight to the biologic behavior of disease. Bone destruction pattern is divided in two types as, bone destruction positive pattern (shows discontinuity of cortex) and bone destruction negative pattern (show expansion, but displays continuity of cortices). CBCT findings in OS can be divided into four types as follows:-

1.osteolytic and bone destruction positive,
2.osteolytic and bone destruction negative,
3.osteogenic and bone destruction positive,
4.osteogenic and bone destruction negative pattern [11].

In present case 3rd type, osteogenic and bone destruction positive pattern is seen.

Different disease shows different type of bone deposition pattern which can be well differentiated on CBCT. Garre's osteomyelitis shows onion skin pattern of periosteal bone deposition. In periostitis there is delicate periosteal elevation with curvilinear band of new bone. Extensive new bone formation in layers parallel to cortex is seen in Ewing's sarcoma and marrow infiltrating diseases like leukemia. Solid homogenous new bone formation without lamellae is seen in benign process such as langerhans cell histiocytosis, healing

fractures, and benign neoplasms. Sunburst appearance and Codman's triangle indicates malignant processes. The differentiation of this bone deposition pattern is well appreciated on CBCT which is important for accurate diagnosis of the diseases [12].

Immunohistochemistry does not play a diagnostic role for OS, since there is no specific marker to distinguish bone matrix from other collagenous counterparts. Osteocalcin is only protein exclusively expressed by the osteoblast and alkaline phosphatase is the enzyme strongly expressed by the all variants of OS. It is helpful in distinguishing primary bone malignancy from others [12].

OS of jaw has tendency to spread locally within the jaw. Surgical exploration for biopsy, extraction of teeth and curettage procedures can cause extraosseous spread of tumor and metastasis. In mandible nerve canal and foramen allow the spread of tumor to extragnathic sites. In maxilla, the tumor can spread to the sinuses, nasal cavity, infratemporal fossa and to other sites since cortices and foramen are thin and can perforate easily. The distant metastasis occurs to lung, brain, other bone, through tumor emboli in veins that either flow downstream to the right side of the heart where they are pumped into and lodged in the capillaries of the lungs or via retrograde flow to the brain or to other bones [13].

Treatment of OS includes surgical excision with involving safe margins, chemotherapy and radiation therapy. Prognosis of OS of jaw is good as compared to the OS of long bone. Factors associated with good prognosis are a small tumor size, absence of nerve involvement, adequate surgical margin, histologically well differentiated lesion, and younger patients without associated retinoblastoma. The 5 years of survival rate with OS of jaw is about 60% to 70%. Therapeutic failure generally occurs due to local recurrence and also due to metastasis to lung followed by metastasis to the brain and then to other bones [13].

Osteosarcoma is a highly malignant jaw bone tumor which has characteristic CBCT features which can be suspected and diagnosed easily by correlating with clinical features. Recently CBCT has been emerged as an advanced imaging tool with less radiation exposure and cost effective as compared to CT and MRI. Bone destruction and bone formation pattern, widening of PDL space, sunburst appearance of bone are the distinguishing features of OS which can be well appreciated on CBCT. These findings will be helpful in differentiating it from other bony lesions like fibrous dysplasia and osteomyelitis. Thus CBCT is an important tool in diagnosis of highly malignant jaw tumor such as OS which can be helpful for early diagnosis and treatment.

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