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RESEARCH ARTICLE

CHONDROSARCOMA FROM OSTEOID OSTEOMA IN MAXILLA- A DIAGNOSTIC DILEMMA

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ABSTRACT

Chondrosarcoma is an infrequent malignant tumor of head and neck with less than 10% of cases occurring in the craniofacial region. Many a times it is a challenge to differentiate its benign or malignant nature. Osteoid osteoma presents with severe localized pain, predominantly nocturnal, relieved with nonsteroidal anti-inflammatory drugs. 80% of osteomas occur in the long bones and less than 1% occur in jaws. Here we present an enigmatous lesion where two tumours with distinct tissue of origin, arose in the same site, started as a harmless benign osteoid osteoma and then transforming into an aggressive sarcomatous lesion.

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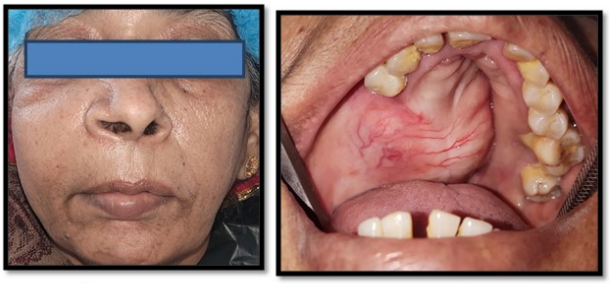
INTRODUCTION

Chondrosarcoma is a malignant neoplasm of cartilaginous origin devoid of tumor osteoid. [1,2]It is an infrequent primary tumor of head and neck with less than 10%of cases occurring in the craniofacial region. Chondrosarcoma are generally seen to occur in the middle age or older adults. The male to female ratio is almost equal with a slight preponderance of males in the ratio of 1.2:1. [1,3,4] Apainless swelling is a typical feature of this lesion. The diagnosis of this tumour is the most arduous one amongst other tumour pathology as it is generally discoverable on radiographs. Many a times it is a challenge to differentiate its benign or malignant nature. Thus, histopathological examination forms the gold standard for final diagnosis. Osteoid Osteoma was distinctively described by Bergstrandin in 1930 and classified by Jaffe in1935. Approximately 80% of osteomasoccur in the long bones- femur (27.33%), tibia (22.1%) spine 10% and less than 1% occur in jaws. [5,6] Most often reported in the 2nd and 3rd decade of life and very rarely occurring in the age of 30 yearsor more havinga male predilection with ratio of 2:1. [3,5-9] Signs of this tumour are of severe localized pain, predominantly nocturnal, relieved by the use of nonsteroidal anti-inflammatory drugs (NSAIDs).[10] Both these tumours are poles apart in their characteristics with no overlapping features clinic-radio-pathologically. Till date, there is no report of either osteoid osteoma converting to malignancy or chondrosarcoma arising out of a benign tumour of bony origin. Here we present an enigmatous lesion where two tumours with distinct tissue of origin, arose in the same site, started as a harmless benign osteoid osteoma and then transforming into an aggressive sarcomatous lesion.

CASE REPORT

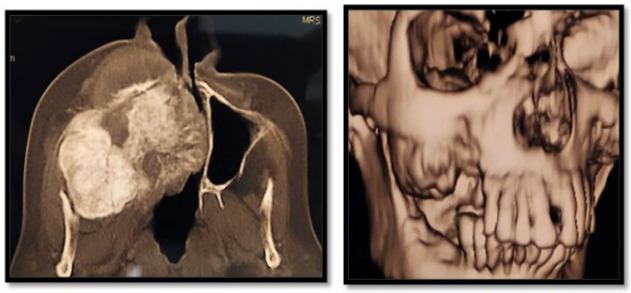
A 65-year-old female patient reported to our department with chief complaint of swelling and mild pain on the right side of face since past 3 months. The patient reported that the swelling had slowly increased in size, initially being painless but subsequently followed by intermittent pain. On extraoral clinical examination, facial symmetry was distorted with a right hemifacial swelling of size around 10x6cm extending antero-posteriorly from midline of face to 1 cm anterior to tragus of right ear and super inferiorlyfrom infraorbital margin to middle of cheek. Swelling in the right-side nasal floor and nasal cavity was seen with obliteration of right nasolabial fold. Intraorally, lesion was approximately 3 x 4 x 4 cm in size obliterating the right maxillary buccal vestibule and pushing the midline of hard palate to the opposite side. The lesion extended antero-posteriorly from canine alveolus area to palatal seal region with missing teeth 15,16 and 17. On palpation, the lesion was soft to hard in consistency, tender, compressible in alveolar region and non-compressible in palatal region and non-reducible. Overlying mucosa was reddish-pink in colour with stretched vascular channels. No ulceration or bleeding was present. (Figure 1a,1b). Radiological examination with CECT neck and face revealed a large expansile bonylesion in the posterolateral wall of right maxillary sinus. It involved right side of nasal cavity, maxilla and hard palate. The lesion measured 3.2 x 3.8 x 4 cm^3in size with associated soft tissue invaginating into right anterior

buccal space region and right masticator space measuring approximately 4.2 x 1.9 cm.



1(a) 1 (b)

Figure 1. Shows clinical features: (a) large swelling seen on right side cheek causing obliteration of nasolabial fold and distortion of nasal cavity (b) intraorally, a single large swelling is noted occupying the palate, obliterating midpalatal raphe and right upper buccal vestibule with intact mucosa. Missing 15, 16 17



2(a) 2(b)

Figure 2. Shows radiological features (a) axial section of CT shows a large ill-defined radiolucent- radiopaque locules of right maxilla, shoving into maxillary sinus and nasal cavity. Sunray pattern is appreciated. Soft tissue shadow is noted plunging to surrounding areas. (b) 3D CT shows the large bony mass on right side of face replacing the maxilla and associated sinus

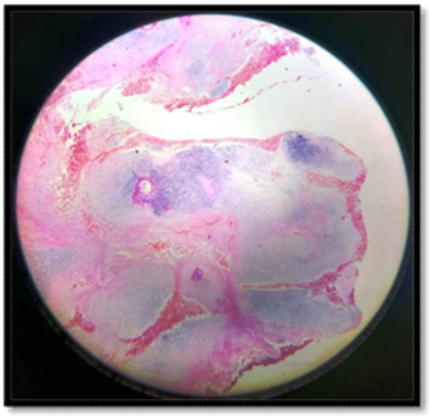


Figure 3: 40x. Histopathologic sections showing areas composed of cartilage with varying degree of maturation and cellularity with typical lacunar formation. Tumour shows a lobular growth pattern separated by a fibrous connective tissue septa.

The lesion was distinctively radio-opaque with a peripheral sun ray type of appearance mesially. Radiolucency of the soft tissue intervened in between as well as surrounded the lesion. (Figure 2a,2b). Multiple enlarged lymph nodes (level 1b) of approximately 9 mm bilaterally were evident. Past medical history revealed patient wasanaemic with Hb-6mg/dl and deranged blood sugar levels. Past dental history revealed that patient had similar tumor like lesion one and half year back at the sameregion for whichpatient underwent incisional biopsy followed by complete excision. Incisional biopsy from the right maxillary tuberosity region with overlying mucosa exhibited stratified squamous epithelium with hyperplasia. Subepithelial stroma was infiltrated with acute and chronic inflammatory cells with marked crushing artefact.

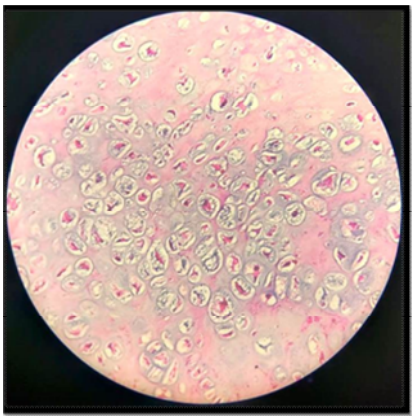


Figure 3b. 400x. Histopathologic sections shows areas composed of chondroid matrix and chondroblasts with subtle variation from normal looking cartilage. Few areas show large plump chondroblasts with varying morphology. Mitosis was rare

Deeper stroma showed metaplastic bone formation and haemorrhagic areas.No evidence of dysplasia or malignancy was evident. The excisional biopsy revealed osseous lesion composed of mature lamellated bony trabeculae and mucosa showing granulation tissue and moderate chronic inflammation. Importantly, all mucosal resected margins were uninvolved and no evidence of malignancy or granuloma formation were noted. Thus, a diagnosis of osteoid osteoma was rendered. However, after one and half yearfollowing surgery, a swelling recurred in the same region and progressively increased in size and symptoms. Patient reported backto the treating clinician after which biopsy was repeatedfrom the same region. Biopsy revealed cellular cartilaginous areas arranged in lobular pattern and mild atypia suggestive of lesion of cellular osseous/cartilaginous origin. Patient was then referred to our hospital for expert opinion and further management. Review of slides showed variably sized lobules of cartilaginous tissue with increased cellularity in chondroid background matrix. Also, chondrocytes with hyperchromatic pleomorphic nuclei and abundant basophilic to vacuolated cytoplasm were present. Occasional mitotic figures were observed. These findings led to the diagnosis in favour of Chondrosarcoma Grade 1. The clinical aggressiveness of the lesion correlated with the histopathological diagnosis. As radiotherapy was not advantageous in low grade chondrosarcoma, patient was scheduled for right hemi maxillectomy followed by fabrication of palatal obturator.

DISCUSSION

The true nature of osteoid osteoma is still unknown. Literature suggests it usually occurs in young adults under 30 years of age which was in contrast to our case as our patient was in her sixth decade of life.[10] Differential diagnosis of osteoid osteoma includes osteomyelitis and osteoblastoma.[5,8] Histologically, osteoid osteomas consist of mature, lamellar bone, or cancellous bone with highly vascular supporting osseous tissue (nidus) surrounded by a distinctive zone of reactive bone formation and abundant fibrofatty marrow between bony trabeculae.[5,6]Our first histopathological report is in accordance with these features. Complete removal of the lesion with the nidus is the treatment of choice. [5,6] Hence, complete excision of tumour was carried out in our patient. To date, no history of malignant transformation of osteoid osteoma has been reported.[10] In our case, after first excisional surgery, cut sections showed tumour free margins histologically, but paradoxically, there was recurrence of the tumour after a year and half. The surprising fact, that benign osteogenic tumours does have potential to turn malignant, however in our literature search we were unable to find any such cases till date where osteoid osteomas have transformed into malignant sarcomas.

Chondrosarcomais a rarity in craniofacial region accounting for 5.76% of all the cases. According to Terezhelmy, the tumour is found in equal frequency in both arches but Huvos reported maxillary predilection with a proportion of 1.75:1.[2] The most common clinical finding is painless swelling, expansion of the bucco-lingual cortex. Rarely lymph node involvement is noted. Our case occurred in a 65-year-old female patient with complaints of a painless swelling over the hard palate, with intact mucosa, consistent with above findings. Premature eruption or exfoliation of teeth is normally found but since this is a recurrent case, patient already had missing posterior molars.[10] Low-grade lesions have a less than 10% risk of metastasis, intermediate-grade lesions have a 10%-50% risk, and high-grade lesion have a 50%-70% risk. The lungs are the main site of metastases., In our patient, CT scan chest did not show any abnormality. The pathogenesis and biologic behaviour are not fully understood. It may be induced by irradiation, from pre-existing Paget's disease, fibrous dysplasia, vestigial cartilaginous rests or de-novo from osseus tissues without the presence of cartilaginous rests. In pre-maxilla and maxilla, the presence of cell rests is highly possible because of the proximity of chondrocranium throughout the fetal development.[1,7] It has been hypothesized that it originates from remnants of embryonic cartilage precursors from nasal and septal developmental region of the maxilla. The radiological presentation of this tumour varies. It may have single or multiple radiolucent areas, opacification of air spaces, a densely calcified bone mass and root resorption, ground glass appearance or a sunburst appearance. In advanced stages, the lesion penetrates the cortical plate and extends into adjacent soft tissues, resulting in a fuzzy peripheral shadow. All these presentations were in accordance with our case.

Histologically, it is composed of fully developed cartilage without tumor osteoid, being directly formed from a sarcomatousstroma. Myxoid changes, calcification and ossification may be seen. In head and neck, the largest percentage of chondrosarcomas has been reported as grade-I. Biopsy report of our case satisfies these criteria with obvious looking multiple lobules of abnormal looking cartilaginous tissue seen within the stroma. All cartilaginoustumours of the jaws, benign or malignant, should be radically excised with a portion of the normal tissue to avoid recurrence. Previous reports states that chondrosarcomasare radio-resistant, thus radiotherapy is not generally recommended as a primary modality and is limited to adjunct to palliative treatment for recurrent lesions. Hence, radical surgery has been planned for presented case here, followed by interim prosthesis. The prognosis is good for low and intermediate grade chondrosarcomas but in jaws it isdisappointingly poor. The cause of death is by direct extension into the base of skull, and through distant metastasis, to lungs and bones. The overall 5-year survival for low grade chondrosarcomas after complete resection is between 55-75%.[1] The reported patient will be kept on regular follow up. Achievement of tumour-free margins is the utmost requirement as the lesion is easily implantable in tissue, leading to rapid growth and invasion. Tumour grade, site and resectability are important prognostic factors.[11]

CONCLUSION

The clinician must always be conscious of the unusual positions and presentation of bony tumours in the maxillofacial region which has to be diligently followed by proper radiographic and histologic correlation for precise diagnosis. This helps in avoidance of misdiagnosis and early multidisciplinary management of complicated lesions. More research is needed to elucidate the nature of progression of bony benign tumours and their conversion rates to malignancy so as to minimise jolt to clinician as well as to the patient.

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