Case Report

Maxillary osteosarcoma masquerading as chronic sinusitis—an intriguing case for surgical pathologists

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Abstract

Osteosarcoma is a highly aggressive malignant bone tumor in which the neoplastic osteoblasts produce osteoid material. This tumor almost exclusively involves the metaphysis of long bones. Involvement of flat bones such as the skull, jaw, and spine are extremely rare. A 22-year-old male patient presented with left-sided nasal obstruction and a painless swelling over the left cheek. On physical examination, a hard, nontender swelling was palpable over the left maxillary region. Computerized tomography (CT) scan of paranasal sinuses revealed that the left maxillary antrum presented an expansile, ill-defined, mixed, radiopaque-radiolucent lesion, invading the cortical bone and adjacent soft tissues. Left hemimaxillectomy was performed including a wide excision of the tumor. On gross examination, a hard bony growth was found in the maxilla. Microscopic examinations revealed histopathological features of osteosarcoma. Osteosarcoma of craniofacial region generally presents in advanced stage in the tertiary-care hospital, mainly, because of rarity of occurrence and lack of awareness. Its clinical features, histological characteristics, and close differential diagnosis are to be kept in mind to avoid late recognition, mutilating surgery, and associated morbidity.

KEY WORDS: Nasal obstruction, osteosarcoma, maxilla

Introduction

Osteosarcoma is a highly aggressive malignant bone tumor almost exclusively involving the metaphysis of long bones of the appendicular skeleton, characterized by neoplastic osteoblasts producing osteoid material.[1] The involvement of nonlong bones such as the skull, jaw, and spine are extremely rare. Only 6% to 10% cases occur in the craniofacial region.[2] It is primarily the tumor of adolescent and young age, as 60% of patients are younger than 25 years of age.[3] Patients with osteosarcoma commonly present with bony mass, pain, and nerve compression symptoms. Nasal obstruction and discharge mimicking sinusitis as a presenting feature is extremely rare.[4]

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We, hereby, present a case of osteosarcoma in maxilla in a 22-year-old male patient creating diagnostic dilemma owing to its rare presentation and location.

Case Reports

A 22-year-old male patient presented in the ENT OPD with the complaints of left-sided nasal obstruction for 1 year; he was provisionally diagnosed to be experiencing sinusitis, but his symptom was not relieved by medications. Over time, a slowly growing painless swelling appeared over his left cheek.

On physical examination, a 5- ×3-cm ill-defined, hard, nontender swelling was palpable over the left maxillary region. No cervical lymphadenopathy was detected. Examination of other systems was unremarkable. Complete blood count, chest X-ray, and abdominal ultrasonography were found to be within normal limits.

Towne's (OM) view plain X-ray of paranasal sinuses revealed a densely calcified and ossified mass in left maxillary antrum. Plain and contrast-enhanced computerized tomography (CT) scan of paranasal sinuses was done, with 5 mm sections both in the axial and coronal planes. Left maxillary antrum showed expansile, ill-defined, mixed,

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radiopaque-radiolucent lesion, invading the cortical bone and adjacent soft tissues resulting in a "sunburst appearance" of periosteum. The lesion extended into frontosuperior margin of left orbit and ethmoid air spaces occluding the left nasal cavity. The mass presented dense amorphous ossifications and showed the heterogeneous uptake of contrast material [Figures 1 and 2].

Biopsy of the growth revealed histopathological features compatible with osteosarcoma. Left hemimaxillectomy was performed including a wide excision of the tumor, and the specimen was sent for histopathological examination. On gross examination, a hard bony growth was found in the maxilla, measuring 5 × 5 × 3 cm. The cross section showed variegated appearance [Figure 3]. Multiple tissue sections were embedded from different parts and were stained with haematoxylin and eosin.

Microscopic examinations showed abundant eosinophilic lace-like osteoid materials lined by highly pleomorphic malignant osteoblasts, confirming the diagnosis of osteosarcoma [Figure 4]. Sections from orbital plate, skin over maxilla, and posterior and lateral margins also revealed the presence of tumor.

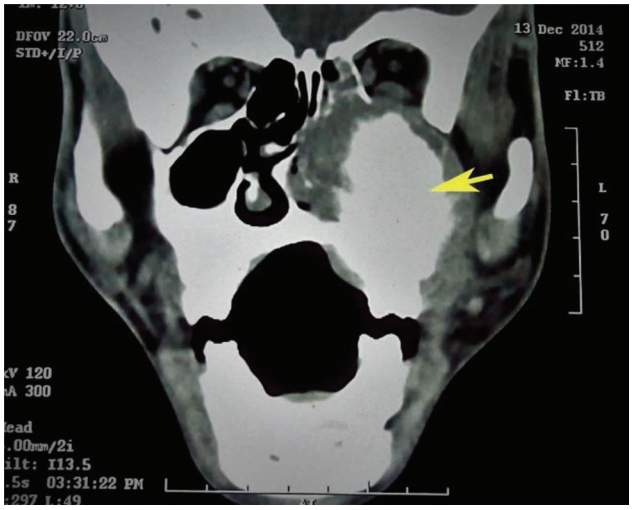

Figure 1: CT scan of paranasal sinuses showing an expansile ill-defined mixed radiopaque-radiolucent lesion in the left maxillary antrum.

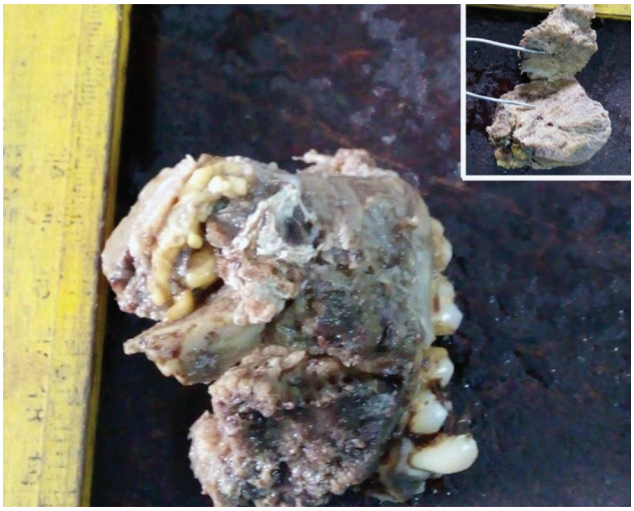

Figure 3: A variegated bony growth found in the maxilla on gross examination (inset, cross section).

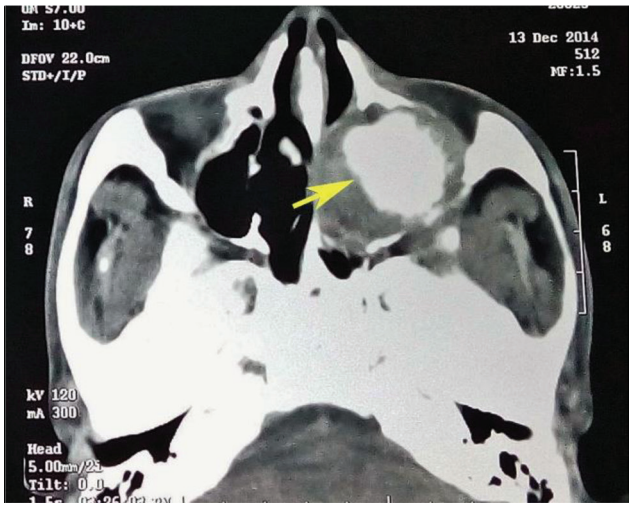

Figure 2: CT scan of paranasal sinuses showing an expansile ill-defined mixed radiopaque-radiolucent lesion invading the cortical bone and adjacent soft tissues.

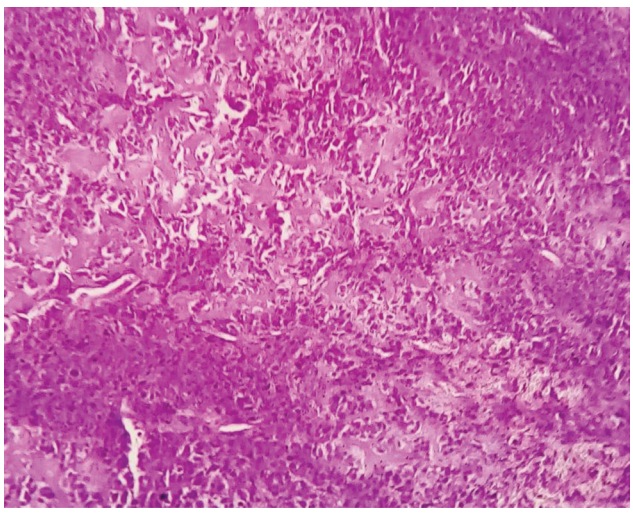

Figure 4: Photomicrograph showing abundant eosinophilic, lace-like, osteoid material lined by highly pleomorphic malignant osteoblasts confirming the diagnosis of osteosarcoma (H and E, 400*x*).

The patient was referred to the Department of Radiotherapy and was on close follow-up then.

Discussion

Osteosarcoma is the most common primary bone tumor, but only 6% to 10% cases occur in the craniofacial region. Among the cases of craniofacial region, the mandible is a comparatively common site, followed by maxilla and skull. When compared with other regions, the craniofacial osteosarcomas are less aggressive and supposed to have local invasion rather than distal metastasis.[5]

Patients with craniofacial osteosarcoma commonly present with bony mass, pain, and nerve compression symptoms. Sinusitis-like features as present in the index case are extremely rare in clinical literatures.

The risk factor for this tumor are preexisting bone diseases or the genetic defects, which are similar to the risk factors for osteosarcoma in other regions. The common preexisting bone diseases that can lead to osteosarcoma are Paget's disease, fibrous dysplasia, chronic osteomyelitis, giant cell tumor, and so on.[6] Our patient did not reveal any of these conditions.

Radiological findings of the craniofacial lesions are nonspecific. They can present as an osteosclerotic/mixed/osteolytic lesion. X-ray must be followed by CT scan. Nowadays, CT scan plays an important role, as it can identify the exact extent of the mass, bony erosion, soft-tissue infiltration, and degree of ossification.[7] The CT findings in our case showed a mixed density lesion, which eroded bone and adjacent soft tissue and invaded the orbit.

Reported craniofacial cases show an equal incidence of osteoblastic, chondroblastic, and fibroblastic types.[8] In our case, the microscopic features were of osteoblastic osteosarcoma.

The ideal treatment for this tumor is complete resection. Total maxillectomy is the recommended surgical procedure. Excision with clear surgical margin is prognostically better and will result in a longer survival.[9] But, in craniofacial region, it is almost impractical to have all surgical margins clear because of the close proximity to the vital structures. Adjuvant treatments must be considered for total complete eradication of the tumor and to prevent recurrence.[10] In our case, the skin over maxilla and the posterior and lateral margins were involved by the tumor. Currently, the patient is receiving adjuvant therapy.

Conclusion

In conclusion, it can be said that osteosarcoma of craniofacial region generally presents in the advanced stage in the tertiary-care hospital, mainly, because of rarity of occurrence and lack of awareness among the treating physicians. Our case is unique on account of its rare presenting features and unusual site of occurrence.

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