Recurrent Mandibular Osteosarcoma with Soft Tissue Extension: A Case Report

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ABSTRACT:
Osteosarcomas are aggressive neoplasms that originate from bone. They are frequently encountered in long bones of extremities with small incidence and rare occurrence in jaws. Twenty-eight-year-old male patient presented with recurrent osteosarcoma of mandible within one year after resection with adjuvant chemotherapy. Swelling, pain, and difficulty with eating were chief complaints. OPG was obtained and revealed characteristic sunburst lesion invading into soft tissue from mandibular body. Hemimandibulectomy was performed and specimen was received by histopathology department at Dow International Medical and Dental College at Ojha campus, Karachi for evaluation. The diagnosis of chondroid variant of mandibular osteosarcomawas made and surgical margins were evaluated for safety.

KEYWORDS: Osteosarcoma, mandible, bone tumor, sunburst lesion, recurrence.


INTRODUCTION
Despite small incidence (3.8 per million in men and 2.8 per million in women) osteosarcoma is the second most frequently documented primary skeletal malignancy after multiple myeloma1. The tumor usually manifests in long bones of extremities with rare (6-9% of all osteosarcomas) occurrence in jaws2. Incidence of osteosarcomas in jaw bones has been reported to be 0.7 per million with slight male predominance (male to female ratio 1.5:1)2,3. Osteosarcomas occurring in jaws are relatively less aggressive than those that take birth in long bones, recurrence after excision, however, is a significant complication4. We present a case of recurrent mandibular osteosarcoma in a 28-year-old male patient that was diagnosed and reported by histopathology department at Dow International Medical and Dental College, Ojha campus, Karachi.

Three variants are described on the basis of histological picture; osteoblastic, chondroblastic, and fibroblastic osteosarcomas. The osteoblastic variant, constituting 60% of jaw lesions, is typified by disorderly arranged malignant osteoblasts with significant pleomorphism and large deep staining nuclei that deposits variable amount of osteoid matrix. Fibroblastic variant is markedly cellular, exhibiting fibroblastic proliferation along with atypical spindle cells and osteoid deposition5. The chondroblastic variant, on the other hand, is characterized by atypical chondroid areas comprising of binucleate cells exhibiting hyperchromatism with prominent nucleoli as was observed in our case6.

CASE REPORT
The patient was a 28-year-old male who reported with primary complaint of pain and swelling in lower border of
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left side mandible associated with feeding difficulty. Detailed history was acquired and revealed the patient was diagnosed and treated for osteosarcoma one year back. An orthopantomogram (OPG) (Fig. 1) was obtained and revealed mixed radiopaque and radiolucent lesion with hallmark sunburst appearance characteristic of osteosarcoma involving the body of left side mandible up to the angle but not crossing the midline. Widening of the periodontal (PDL) space and loss of lamina dura was also appreciated around left mandibular canine and premolars.

**Fig. (1).** OPG showing characteristic sunburst lesion.

The hematological and biochemical tests were all within normal limits. Serological tests performed for hepatitis B and C were non-reactive.

Hemimandibulectomy was done and specimen was received by histopathology department at Dow International Medical and Dental College, Ojha campus, Karachi for evaluation.

Histopathologic examination displayed malignant neoplasm composed of spindle shaped cells displaying moderate to marked nuclear pleomorphism and hyperchromasia. Malignant osteoid formation was noted in some places. Presence of chondroid areas was also appreciated, hence the diagnosis of chondroblastic variant of osteosarcoma was made after consideration of chondrosarcoma as differential diagnosis (Picture 1 and 2).

**DISCUSSION**

Osteosarcomas or osteogenic sarcomas are most commonly reported primary bone tumors in adolescents/young adults characterized through deposition of osteoid or bone formation by neoplastic cells. Overall incidence is small encompassing merely ~0.2% of all malignancies. Occurrence in jaws is even rarer (6-9% of all osteosarcomas) with relatively greater prevalence reported in older age group (34-36 years). A broad spectrum of clinical, histological, and radiographic presentations is recognized and hamper the process of reaching a conclusive diagnosis.

Hallmark clinical manifestation includes swelling and pain, both observed in our case along with difficulty in feeding.

Owing to overlapping radiographic and histological features, it is difficult to differentiate between chondrosarcomas and osteosarcomas with chondroid differentiation. In our case, presence of large chondroid areas also posed same problem, but the characteristic sunburst radiographic picture as well as osteoid deposition histopathologically aided to rule out chondrosarcoma. Radiography is an indispensable tool in diagnosis of mandibular osteosarcoma whereby widening of the PDL space around teeth in involved area, sunburst appearance, and widening of mandibular canal are pathognomonic. Yen et al., recognized that origin from metaphysis, bone-forming matrix, marked periosteal reaction and patient from younger age group favor diagnosis of chondroblastic variant of osteosarcoma over chondrosarcoma.

Fibrous dysplasia, osteomyelitis, osteoma, myositis ossificans, and cemento-osseous dysplasias are commonly occurring lesions in mandible that may resemble osteosarcoma on radiological picture. Histopathology remains gold standard to differentiate osteosarcoma from these differentials.

Multiple incidences of recurrence have been reported in association with osteosarcoma of jaws. Two such cases were observed with chondroblastic variant of osteosarcoma, similar to our case. Wide variation exists with respect to time span observed before appearance of recurrence from 3 years to as short as 1 month. In reported case, recurrence was recorded within one year, intermediate between reported durations observed before appearance of recurrent tumor.

Wide excision with clear surgical margins are mainstay of eradication and prevention of recurrence. Excision without disease-free margins renders radiation therapy inadequate. The recurrence in presented case was recorded within one year of excision with adjuvant chemotherapy. Without resection disease progression can be fatal with 5-year survival rate of 62%.

**CONCLUSION**

Osteosarcomas are challenging neoplasms for both, histopathologists from a diagnostic point of view, and surgeon because of the risk for recurrence. We have reported
a case of fairly rapid recurrence (within a span of one year) to highlight the significance of wide clear surgical margins in management of osteosarcomas of jaws.

CONFLICT OF INTEREST

Declared none.

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Declared none.

REFERENCES