

Case Report

Metastatic Osteosarcoma to the Maxilla

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ABSTRACT

We present a case of successfully treated primary osteosarcoma (OS) of the tibia with an isolated maxillary metastasis, which is an extremely rare entity. Once histopathologically proved, a total maxillectomy with adjuvant chemo-radiation was carried out. Local recurrence and distant metastasis have been negative for

the past year. Surgery is the gold standard of treatment in operable cases. Adjuvant treatment is required to prevent local recurrence and distant metastases. A multi-modality therapeutic approach is essential in treating these patients.

KEYWORDS: chemo-radiation, maxilla, metastasis, osteosarcoma, surgery

INTRODUCTION

Primary osteosarcoma (OS) is the most common malignant tumor of bone, evolving from bone forming cells. The most common location is the metaphyseal region. OS has a strong tendency for the development of lung metastases^[1,2]. Though most deaths have occurred with uncontrolled disseminated disease, recent advances in surgery (thoracoscopy and thoracotomy with resection of pulmonary metastases), combination chemotherapy and radiotherapy have remarkably improved the survival rates^[1].

OS of the maxillofacial region (primary or metastatic) is rare^[1]. Approximately 7% of all primary OS arise in the jaw bones^[3]. The mandible is more commonly involved than the maxilla (1.5:1)^[3,4]. It is this rare event that has prompted the following report of a metastatic maxillary tumor from a successfully treated OS of the tibia.

CASE REPORT

A 18-year-old male was successfully treated for OS of right tibia with neo-adjuvant chemotherapy (three cycles of methotrexate and cisplatin) and a local resection with limb reconstruction (with Ilizarov external fixator). Six months later he presented to us with complaints of pain and a swelling of the palate near the region of the upper right third molar of 15 days' duration. It bled easily and caused difficulty in mastication. There was no history of tooth extraction in this area. A detailed history for any distant metastasis (lung, liver, lymphnodes, and bones) was negative.

Intraoral examination revealed a tumor mass on the palatine mucosa near the region of the upper right third molar (Fig. 1). The lesion was around 3 x 4 cm in diameter. The surface of the tumor was rough, bled easily on probing, relatively non-tender and the surrounding tissue appeared to be infiltrated. The teeth in this region were predominantly healthy with no dental caries. This mass did not appear to be dental in origin and hence a biopsy was performed. The histopathology revealed a tumor composed of sheets of oval cells with hyperchromatic nuclei and eosinophilic cytoplasm. Areas of osteoid and woven bony spicules were seen and thus confirmed the diagnosis of OS (Fig. 2 & 3). An intraoral periapical radiograph of right upper molar region was normal. A chest X-ray, computed tomography (CT) scan of the chest and a bone scan were negative for distant metastasis.

A total maxillectomy with adjuvant chemo-radiation was carried out. The patient has been kept under close follow-up for local recurrence and distant metastasis, which have been negative for the past year.

DISCUSSION

Metastatic tumors to the oral region are far less common than primary oral lesions but are no less important. One percent of all oral malignancies represent metastatic foci. A metastatic lesion of the oral cavity may be the first evidence of malignant disease elsewhere. In about 33% of patients, oral secondary tumors are the initial indicators of the existence of the primary tumors. The malignant

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Fig 1: Intraoral photograph showing a tumor mass near the region of upper right third molar.

tumor that most commonly metastasises to the oral region is breast cancer. Ninety percent of all oral metastatic tumors occur in the jaws, of which 72% occur in the mandible. The maxilla is the site of metastases in only 18% of patients^[5].

Primary maxillary OS involving the alveolar ridge or antrum^[3,4] is a well-known entity and 56 cases have been reported so far in literature^[6]. But as far as metastatic maxillary OS is concerned, to our knowledge, only one case has been reported, in which the primary was the femur^[7]. This is the second case to be reported and the metastasis was from an OS of the tibia. The criteria for considering a malignant neoplasm to be metastatic are that firstly there must be a histologically verified primary and secondly the secondary lesion must be histologically the same as the primary^[8].

In contrast to our patient, majority of the patients with oral metastases are between 40 and 60 years of age^[5]. OS is a disease which occurs in early age and has a high malignant potential to metastasise usually to the lungs and liver. Distant metastases from a primary OS occurs via the blood stream and lymphatics. Aisenberg^[9], Cataldo *et al*^[10] and Shapiro *et al*^[11] have reported cases of mandibular metastases of OS. A maxillary metastasis is extremely rare and when it occurs, the premolar-molar area is commonly affected^[8], as in our patient. Metastases to the jaws without lung involvement are rare. Batson's plexus has been mentioned as a possible route of spread, thus explaining why in some cases of metastases, the lungs are not involved, as the blood has not been filtered through the pulmonary bed before reaching the head and neck^[8]. This may have happened in our case.

The symptomatology of pain, swelling, paresthesia, loosening of teeth, failure of extraction sites to heal, and enlargement or deformity of the bone are commonly present. The first two symptoms were present in our patient. X-rays

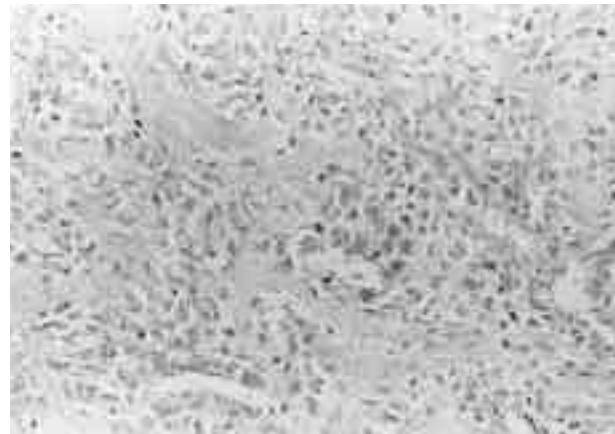


Fig 2: Photomicrograph showing areas of osteoid, sheets of oval cells with hyperchromatic nuclei (H&E x 20).

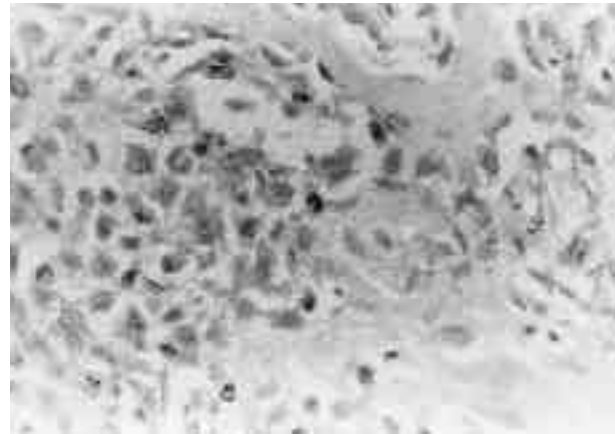


Fig 3: Photomicrograph showing pleomorphism of cells and nuclei with mitotic figures interspersed with osteoid areas (H&E x 40).

usually show radiolucent areas and destruction of cortical plate, but in early and small lesions these findings might be absent^[12], as in our case. However, no characteristic oral manifestations of metastatic OS can be identified because of its rare incidence. Consideration should be given to the possibility of oral metastases in patients with known primary malignant disease and biopsy is essential for establishing the diagnosis^[1]. A rapidly growing inflammatory lesion or a malignant tumour should be suspected, if destruction of the cortex is present^[5]. CT scan and magnetic resonance imaging (MRI) are valuable adjuncts to plain films for determining the extent of the disease^[13].

A multimodality therapeutic approach is required for treating these patients. Surgery (radical resection) is the mainstay and gold standard of treatment in operable cases. Although radical surgery provides long-term disease-free survival for patients with primary disease,^[1,14] treatment modalities for oral metastases have not been established because of the extremely rare incidence, resulting in poor patient prognosis. Resection of oral metastases improves oral function (mastication)^[1]. Adjuvant chemo-radiation seems to be a must for bulky disease, to prevent local

recurrence and systemic metastases^[15]. Intensive combination chemotherapy with high dose methotrexate, doxorubicin, cisplatin, ifosfamide and etoposide has been commonly recognised as the most potent chemotherapeutic protocol^[1]. Neo-adjuvant chemo-radiation helps in shrinking the tumor size^[6]. Inoperable lesions are better treated palliatively by chemo-radiation. Massive bleed from maxillary metastases warrants maxillary artery ligation.

The over all 5-year survival rate for primary OS of jaws varies from 30% to 40% with survival up to 80% reported for patients undergoing early radical resection^[4]. Though the prognosis in maxillary metastases of OS is guarded, it is difficult to ascertain the same, as this is the second case report of isolated maxillary metastasis of OS.

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