Metastatic osteosarcoma to the maxilla: A case report and a review of the literature

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A patient with a successfully treated primary osteosarcoma of the tibia experienced an isolated maxillary metastasis, which is an extremely rare entity. Once the tumor was confirmed histopathologically, a total maxillectomy and adjuvant chemoradiation therapy were carried out. Here has been no local recurrence or distant metastasis for 1 year. A multimodality therapeutic approach is essential when these patients are treated. Surgery is the gold standard of treatment in operable cases. Adjuvant treatment is required to prevent local recurrence and distant metastasis. (Quintessence Int 2002;33:397-399)

Key words: chemoradiation, maxilla, metastasis, osteosarcoma, surgery

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

Osteosarcoma of the maxillofacial region (primary or metastatic) is rare.1 Approximately 7% of all primary OSs arise in the jawbones.3 The mandible is more commonly involved than the maxilla (1.5:1).1 It is the rare event that has stimulated the following report of a metastatic maxillary tumor from a successfully treated OS of the tibia.

CASE REPORT

A 18-year-old man presented with complaints of pain and swelling of the palate, near the maxillary right third molar; the swelling was of 15 days' duration. About 6 months earlier, he had been successfully treated for OS of the right tibia with neoadjuvant chemotherapy (three cycles of methotrexate and cisplatin) and a local resection with limb reconstruction (with an Ilizarov external fixator). The swelling 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, lymph nodes, and bones) was negative.

Intraoral examination revealed a tumor mass on the palatine mucosa near the region of the maxillary right third molar (Fig 1). The lesion was about 3 X 4 cm in diameter. The surface of the tumor was rough, bled easily on probing, and relatively nontender, and the surrounding tissue appeared to be infiltrated. The teeth in this region were predominantly healthy and exhibited no dental caries. This mass did not appear to be of dental origin, and hence it was decided to biopsy the tumor.

A wedge biopsy revealed a tumor composed of sheets of oval cells with hyperchromatic nuclei and eosinophilic cytoplasm. Areas of osteoid and woven bony spicules confirmed the diagnosis of OS (Figs 2 and 3). The findings on the intraoral periapical radiograph of the maxillary right molar region were normal. A chest radiograph, computed tomography scan of the chest, and a bone scan revealed no distant metastases.
A total maxillectomy and adjuvant chemoradiation therapy were carried out. The patient has been kept under close follow-up. There has been no local recurrence or distant metastasis for the past year.

**DISCUSSION**

Metastatic tumors to the oral region are far less common than are primary oral lesions but are no less important. About 10% 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 330,000 patients, oral secondary tumors are the initial indicators of the existence of the primary tumors. The malignant tumor that most commonly metastasizes to the oral region is breast cancer. Approximately 90% of all oral metastatic tumors occur in the jaws, and of these, 720,000 occur in the mandible. The maxilla is the site of metastases in 180,000 patients.

Primary maxillary OS involving the alveolar ridge or antrum is a well-known entity, and 56 cases have been reported in the literature. However, only one case of metastatic maxillary OS has been reported so far; in that case, the primary OS affected the femur. The present case report it; the second regarding metastatic maxillary OS, but the first to describe a metastasis from OS of the tibia. There are two criteria for considering a malignant neoplasm to be metastatic: (1) There must be a histologically verified primary tumor, and (2) the secondary lesion must be histologically the same as the primary.

In contrast to this patient, the majority of patients with oral metastases are between 40 and 60 years of age. However, because OS is an early-age disease, all patients have a high malignant potential to spread, it usually metastasizes to the lungs and liver. Distant metastases from a primary OS occurs via the bloodstream and lymphatic system. Aisenberg and Jmman,9 Cataldo et al,10 and Shapiro et al11 have reported cases of mandibular metastases of OS. A maxillary metastasis is extremely rare; when it occurs, the premolar-molar area is commonly affected, as in the present patient. Metastasis to the jaws without lung involvement is rare. Batson's plexus has been mentioned as a possible route of spread, thus explaining why, in some cases of metastases, the lung is not involved, because the blood has not been filtered through the pulmonary bed before reaching the head and neck. This may have happened in the present case.

Pain, swelling, paresthesia, loosening of teeth, failure of extraction sites to heal, and enlargement or deformity of the bone are commonly present, and the first two were found in this patient. Radiographs usually reveal radiolucent areas and destruction of the corticosteroid.
but in early and small lesions these findings
may be a sensitive to the recent case. However, no
correlation of oral manifestations of metastatic OS can
be identified, because of its rare incidence.

Consideration should be given to the possibility of
tatal metastases in patients with known primary malign-
ant disease. Biopsy is essential to establish the
diagnosis. A rapidly growing inflammatory lesion or a
malignant tumor should be suspected if destruction of
the cortex is present. 5 Computed tomography and mag-
netic resonance imaging are valuable adjuncts to plain
radiographs 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 dis-
case, multimodal treatment modalities for oral metastases have
now been established because of their extremely rare
presentation, resulting in poor prognoses. Resection of
oral metastases improves oral function (mastication). 1

Adjuvant chemoradiation seems to be a must for bulky
disease. Intensive combination chemotherapy with high-dose methotrexate, doxorubicin, cisplatin, ifosfamide,
and etoposide has been commonly recognized as the most potent chemotherapeutic protocol. 1
Neoadjuvant chemoradiation helps to shrink the mmor.
Inoperable lesions are better palliated by d11cmoradiation. Massive bleeding from maxillary mela
tasis warrants ligation of the maxillary artery.

Therapy rates 11 5-year survival rate for primary OS of
the jaws varies from 30% to 40%, and survival rates of
up to 90% have been reported for patients undergoing
early radical resection. Although the prognosis for
patients with maxillary metastases of OS is guarded,
their true prospects are difficult to ascertain because
this is only the second case report of an isolated max-
illary metastasis OS.

CONCLUSION

A patient with a successfully treated primary osteosar-
coma of the tibia experienced an isolated maxillary
mela. A total maxillectomy and adjuvant
chemoradiation therapy were carried out. There has
been no local recurrence or distant metastasis for 1
year. A multimodality therapeutic approach to treatment
of metastatic osteosarcoma is essential.

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