

CASE REPORT

## Metastatic Osteosarcoma with Intratumoral Hemorrhage at Skull Base

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### ABSTRACT

*Osteosarcoma is the most common malignant bone tumor in adolescents. Brain metastases are rare and initial presenting symptoms appear in the later stages. A 20-year-old female presented with a two-month history of headache, diplopia and deafness in the right ear. She had a history of surgery for left leg osteosarcoma two years before. We describe a case of metastatic osteosarcoma involving the skull base and presenting with cranial nerve palsies.*

**Key words:** Osteosarcoma, Metastasis, Skull base, Hemorrhage

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### ÖZET

#### Kafa Tabanında Tümör İçi Kanama ile Birlikte Metastatik Osteosarkoma

*Osteosarkom, ergenlerde en sık görülen kötü huylu kemik tümörüdür. Beyin metastazı nadirdir başlangıç belirtileri ileriki aşamalarda görünür. Yirmi yaşındaki kadın, iki aylık bir geçmişi olan baş ağrısı, çift görme ve sağ kulakta sağırılık ile başvurdu. İki yıl önce, sol bacakta osteosarkom için cerrahi bir geçmişi vardı. Biz kraniyal sinir felciyle başvuran ve kafa tabanını kapsayan metastatik osteosarkomlu olgu sunuyoruz.*

**Anahtar kelimeler:** Osteosarkom, Metastaz, Kafa tabanı, Kanama

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## INTRODUCTION

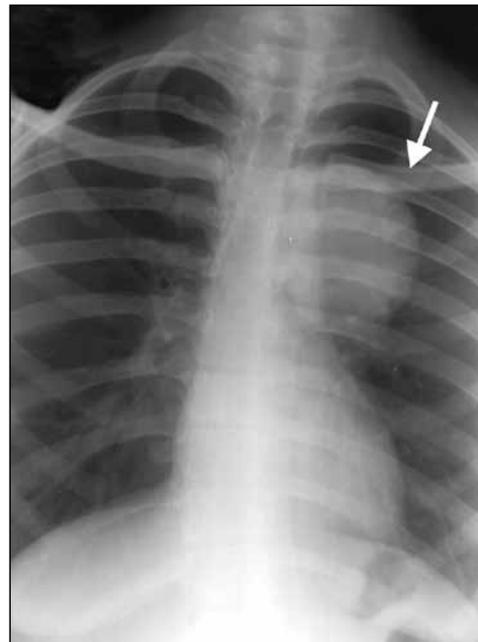
Osteosarcoma is a highly malignant and the most common primary tumor in the bones. It is especially seen in adults and localized at the proximal tibia and distal femur<sup>[1-3]</sup>. It metastasizes easily via a hematogenous route to the lungs, bones and brain, and to the skull base, which is quite rare. Osteosarcomas must be differentiated from other regional, primary and metastatic tumors during clinical and radiological diagnosis<sup>[2-4]</sup>. We report a case of metastatic intracranial osteosarcoma with intratumoral hemorrhage at the skull base.



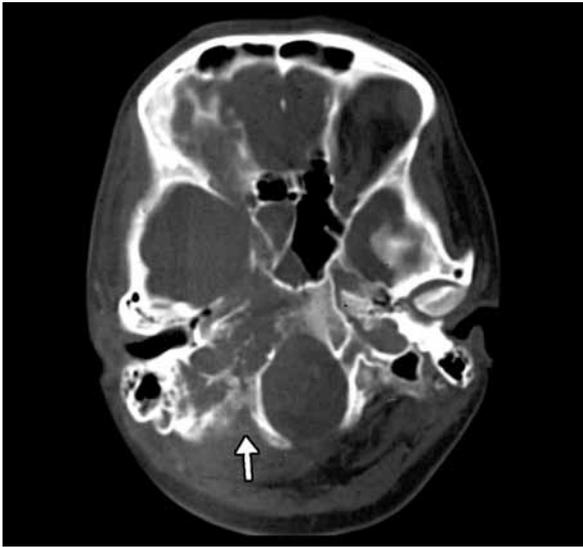
**Figure 1. (A)** X-ray showing tumor of the left tibia. **(B)** Appearance of the chondroblastic components of the osteosarcoma on histopathological examination.

## CASE REPORT

A 20-year-old female patient was admitted to the hospital with a six-week history of headache, diplopia, dysphasia, deficiency in taste of foods, and deafness in the right ear. Her medical history revealed that she had undergone an upper knee left leg amputation two years before, and the pathological diagnosis was osteoblastic and chondroblastic cell type osteosarcoma (Figure 1 A,B). Despite radiotherapy and chemotherapy, solitary pulmonary metastases occurred (Figure 2). Her general condition worsened. Neurological examination showed anisocoria with right pupillary dilatation, diplopia, restriction of right lateral gaze, and right facial hypoesthesia. Deafness in the right ear, loss of taste, dysphasia and dysarthria, weakness and deviation to the left of the soft palate and to the right in the tongue, and difficulty in tongue movement were present. Weakness of the sternocleidomastoid muscle on the left side, inability to hold the head straight, and hemiparesia on the left and extensor response on the right plantar reflex were also determined. An axial computerized tomography (CT) scan demonstrated a tumoral lesion on the right with destruction of the skull base and extension to the bulbar region, hyperdense in accordance with hemorrhage (Figure 3). Magnetic resonance imaging (MRI) revealed that the results of the CT scan further proved a metastatic tumoral lesion and subacute phase on the intratumoral hemorrhage at the bulbar region



**Figure 2.** Pulmonary involvement is seen in the X-ray.



**Figure 3. Contrast-enhanced cranial CT showing the tumor spreading to the skull base.**

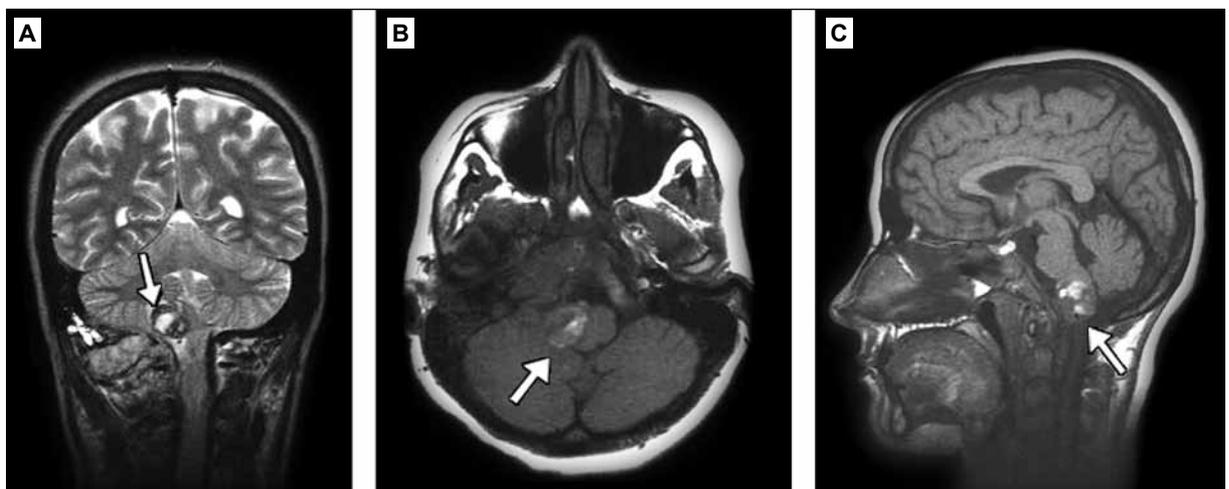
(Figure 4A-C). MRI revealed isointensity with muscles on T1-weighted (W) and hyperintensity with the basis cranii, clivus and dorsum sella on T2-W images, destruction of occipital and temporal bone on the right side and destruction of the foramen magnum, and medulla oblongata compression from the right anterolaterally. The MRI also showed the late subacute hemorrhage within the tumoral lesion in the bulbar region that became hyperintense on T1-W and T2-W images. After an intravenous injection of gadolinium, the pattern of the tumor enhancement was diffuse and homogeneous (Figure 5A,B). In a biochemical examination,

the serum alkaline phosphatase level was three-times elevated. The patient was discharged from the hospital since she and her family refused any treatment.

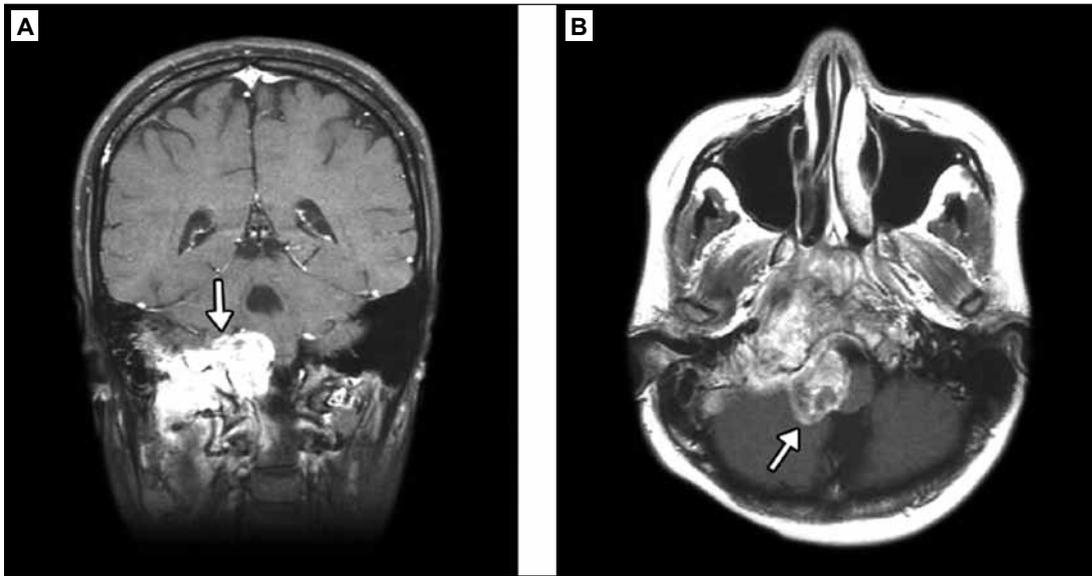
## DISCUSSION

Osteosarcomas are the most common and highly malignant bone tumors. They are derived from mesenchymal cells and are seen in adolescents. These tumors have a tendency for skeletal region metaphases of long bones. Facial bones, skull bones and brain metastases are rarely seen<sup>[1-3]</sup>. Hematogenous metastasis is common. Lungs and other bone metastases are the most frequently occurring types, while localization in the brain and skull base are rare. The central nervous system is usually protected by the presence of the blood brain barrier<sup>[2-4]</sup>. Cranial tomography and MRI exams are useful for the localization of these tumors. Biopsy must be performed for definitive diagnosis and differential diagnosis<sup>[4-6]</sup>.

Various publications have been reported in the literature concerning the metastasis of osteosarcomas, with localizations in various parts of cranial and facial bones, involvement of cranial nerves, and sometimes causing varying neurological damage<sup>[3-5]</sup>. While Caron et al. detected cranial involvement in 11 of 43 patients who were followed with the diagnosis of osteosarcoma, Lam et al. presented a case of multicentric sclerosing osteosarcoma in a 16-year-old girl that resulted in death due to the rapid progression of cranial nerve palsy<sup>[6,7]</sup>. Bindal et al. detected osteosarcoma as the primary tumor in 6 of 21 patients with brain



**Figure 4. Unenhanced (A) coronal and (B) axial cranial MRI showing the hemorrhagic appearance in the bulb in the subacute phase. (C) Unenhanced cranial MRI with sagittal section showing hemorrhagic appearance in the bulb.**



**Figure 5. Contrast-enhanced (A) coronal and (B) axial cranial MRI showing invasion of the skull base.**

metastasis of the sarcoma, and reported that recurrence was seen within two to four months following surgery<sup>[8]</sup>. Sato et al. published a case of unilateral osteosarcoma with neurologic symptoms due to the compression of the IX<sup>th</sup>, X<sup>th</sup> and XI<sup>th</sup> cranial nerves and with the involvement of craniofacial bones and an aggressive course<sup>[9]</sup>. Although all of these cases were followed after surgery, chemotherapy or radiotherapy, the survival rate was found to be very low.

Our patient represents a rapidly progressive case of osteosarcoma compressing the surrounding cranial and spinal nerves due to the skull base metastasis, causing neurological damage and also bleeding in the bulbous; to our knowledge, it is the first case in the literature with these features.

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