Osteoblastoma-like osteosarcoma of the cervical spine: A case study

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ABSTRACT
Osteoblastoma-like osteosarcomas, a less aggressive variant of osteosarcomas, are rare tumors presenting a clinical and histopathological diagnostic challenge. We have found few case reports of this tumor in the literature. In this case, we describe an osteoblastoma-like osteosarcoma of the cervical spine in a 16-year-old patient. CT-guided bone biopsy showed a lesion with a permeated growth pattern, which led to the suspicion of an osteoblastoma-like osteosarcoma. A total *en bloc* spondylectomy was performed through a dual approach. We present the clinical case, the diagnostic sequence, the surgical approach and 10-year follow-up results.
Key words: Osteoblastoma-like osteosarcoma; aggressive osteoblastoma; spondylectomy; tumor; cervical spine.
Level of evidence: IV

Osteosarcoma "tipo osteoblastoma" de columna cervical. A propósito de un caso

RESUMEN
El osteosarcoma "tipo osteoblastoma", una variante menos agresiva del osteosarcoma, es una enfermedad poco frecuente y representa un desafío diagnóstico tanto clínico como histopatológico. Se han publicado escasos reportes de casos de este tumor. Presentamos a un paciente de 16 años con un osteosarcoma "tipo osteoblastoma" localizado en la columna cervical. La biopsia ósea bajo tomografía reveló una lesión con un patrón de crecimiento permeativo, con sospecha de osteosarcoma "tipo osteoblastoma". Se realizó una espondilectomía total en bloque mediante un doble abordaje. Se describen el caso clínico, la secuencia diagnóstica, la técnica quirúrgica y el seguimiento a 10 años.
Palabras clave: Osteosarcoma tipo osteoblastoma; osteoblastoma agresivo; espondilectomía; tumor; columna cervical.
Nivel de Evidencia: IV

INTRODUCTION

Primary spinal bone tumors are rare lesions accounting for 90% of metastatic tumors.[1] The most common primary tumors are osteoid osteomas and osteoblastomas, with an incidence rate of 10% in the spine.[2-4]

Osteoblastomas and osteosarcomas can be difficult to tell apart both clinically and histopathologically, and the presence of conditions that can be mistaken for them is a challenge for diagnosis and treatment of these tumors.[5] Osteoblastoma-like osteosarcomas are a rare type of tumor accounting for 1.1% of osteosarcomas. This secondary type of osteosarcomas resembles an osteoblastoma histologically speaking, because it fabricates the same type of microtrabecular bone covered by osteoblasts.[6] It is extremely important to make an accurate diagnosis due to the recurrence rate and the potential risk of metastasis of these tumors, unlike osteoblastomas.[7]

We present the case of a young patient with an osteoblastoma-like osteosarcoma of the cervical spine, the diagnostic sequence, the surgical approach and 10-year follow-up results.

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CLINICAL CASE

A 16-year-old boy was admitted to our institution with acute post-traumatic cervical non-radiating pain. The patient was referred after a finding on a cervical X-ray. The physical exam was normal.

X-rays of the cervical spine without contrast revealed a radiopaque (blastic) lesion on C5 (Figure 1). Due to this finding, a CT and an MRI were requested in order to further characterize the lesion.

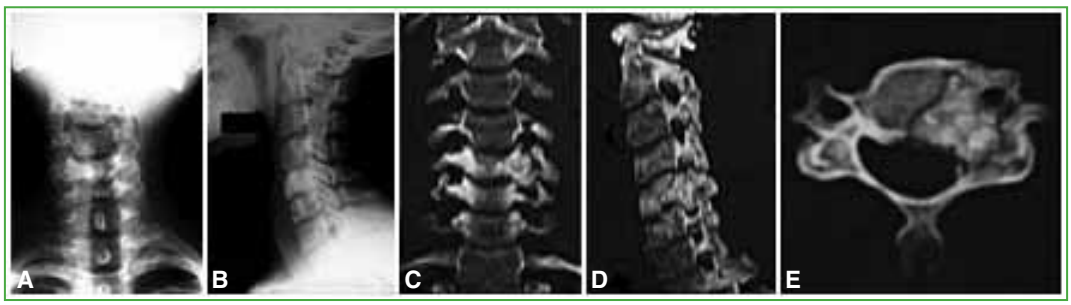

Figure 1. A and B. AP and lateral X-rays of the cervical spine without contrast. A sclerotic bone lesion is observed in C5 vertebral body. **C-E**. CT scan. Well-defined tumor involving the left vertebral body.

The CT showed a sclerotic bone lesion on the left side of the C5 vertebral body, compromising the pedicle and the transverse foramen of the ipsilateral vertebral artery (Figure 2).

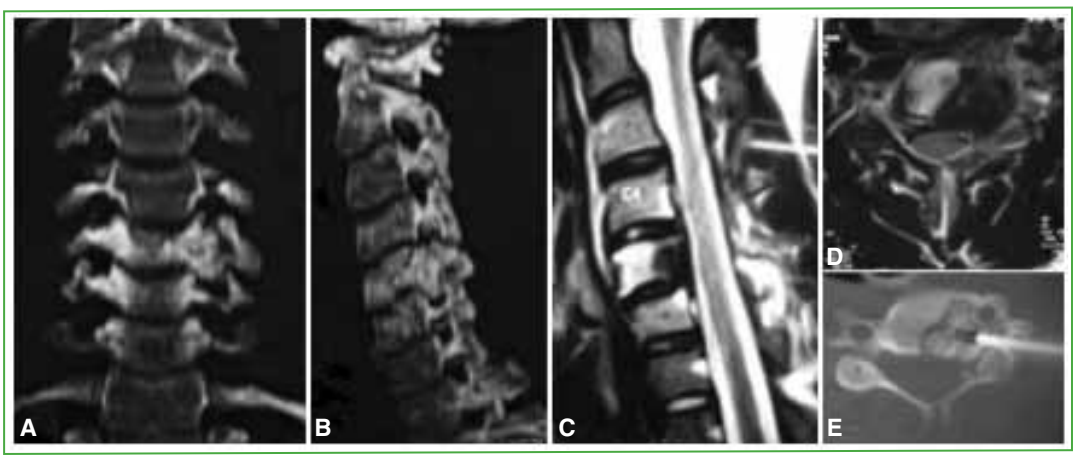

Figure 2. A-D. CT and MRI scans. The tumor can be observed in C5. **E.** CT-guided biopsy.

The MRI showed a hypointense image in T1- and T2-weighted scans on the left side of C5, without involvement of the spinal cord; however, surrounding soft-tissue involvement, a pathognomonic sign of an aggressive tumor, was observed.

A CT-guided percutaneous biopsy was performed. The biopsy showed an osteoblastic tumor with areas of an osteoblastoma-like pattern mixed with a malignant bone-forming tumor.

A tumor compatible with an osteoblastoma-like osteosarcoma was diagnosed. Consequently, a total *en bloc* spondylectomy was performed through a dual approach.

Surgical planning

During the preoperative exam, a compression test of the vertebral artery[8] was performed on the left side to evaluate arterial dominance of the area. In addition, incisions were practiced on a 3D model (Figure 3A) to achieve a better spatial and anatomical appreciation of the tumor.

A total *en bloc* spondylectomy was then carried out by a combined anterior and posterior approach. The first step, with the patient in the ventral decubitus position and through a posterior approach, consisted in ligament and muscle release, identification of C5 under fluoroscopy, and resection of the posterior arch. In addition, posterior C3-C6 instrumentation was performed—lateral mass screw fixation—, in order to provide spinal stability.

In the second step, during the same surgical stage and after placing the patient in the dorsal decubitus position and at 45°, resection of the C5 vertebral body was performed by an anterior *en bloc* spondylectomy, preserving the foramen of the right vertebral artery. The space was filled with a titanium mesh cage and an autologous bone graft inside the cage.

The procedure lasted 340 minutes and the estimated blood loss was 250 mL. Both steps were performed under neurophysiologic monitoring, which included somatosensory and motor evoked potentials. Antibiotic prophylaxis was administered according to the usual protocol proposed by the Infectious Disease Department of our institution.

No significant intraoperative complications were observed. The patient remained in the ICU for 2 days and was discharged after 7 days.

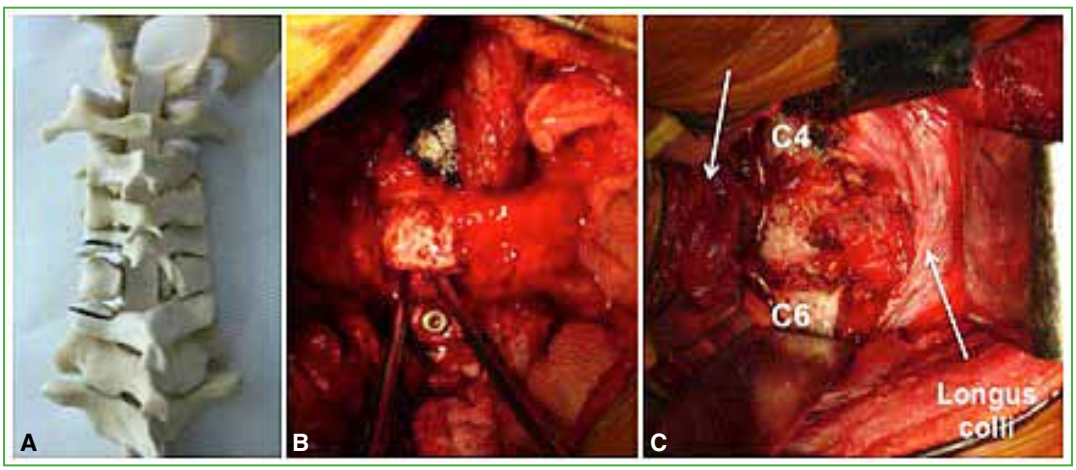

Figure 3. A. Planned spondylectomy. **B and C.** Intraoperative images of *en bloc* resection.

Histopathology

The histopathological examination shows round and oval osteoblastic tumor cells with large nuclei that contain prominent nucleoli, with a minimal osteoid component mitotic activity. In addition, it contains solid cell nests of a non-bone-forming nature. The tumor is permeating entrapped bony trabeculae, which is not the case of osteoblastomas or aggressive osteoblastomas (Figure 4).

Follow up

No surgical complications were detected. After surgery, the patient received adjuvant chemotherapy with methotrexate, ifosfamide and doxorubicin for four weeks.

A chest CT was taken every 3 months during the first 2 years, every 6 months for the following 2 years and once a year after that.

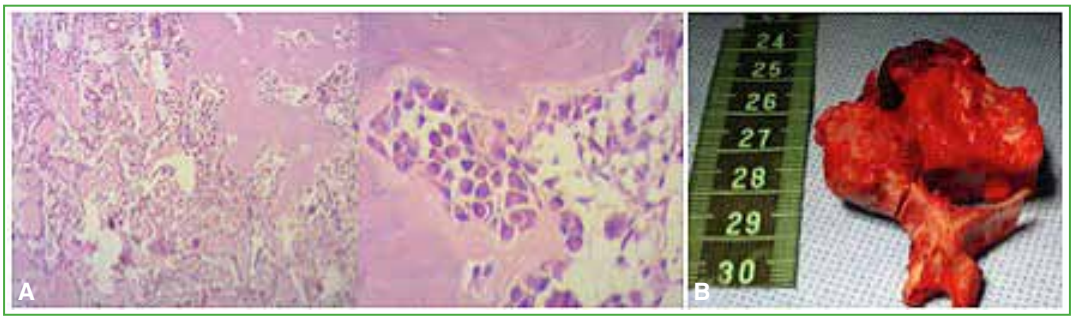

Figure 4. A and B. Microscopic section of the vertebral body showing large active hyperchromatic cells associated with thick strands of bone matrix. **C.** Macroscopic vertebral section.

X-rays were normal during the follow-up, without signs of lack of consolidation or spinal imbalance. At the 10-year follow-up, there was no local recurrence or distant metastasis. In addition, X-rays without contrast showed a solid cervical arthrodesis (Figure 5).

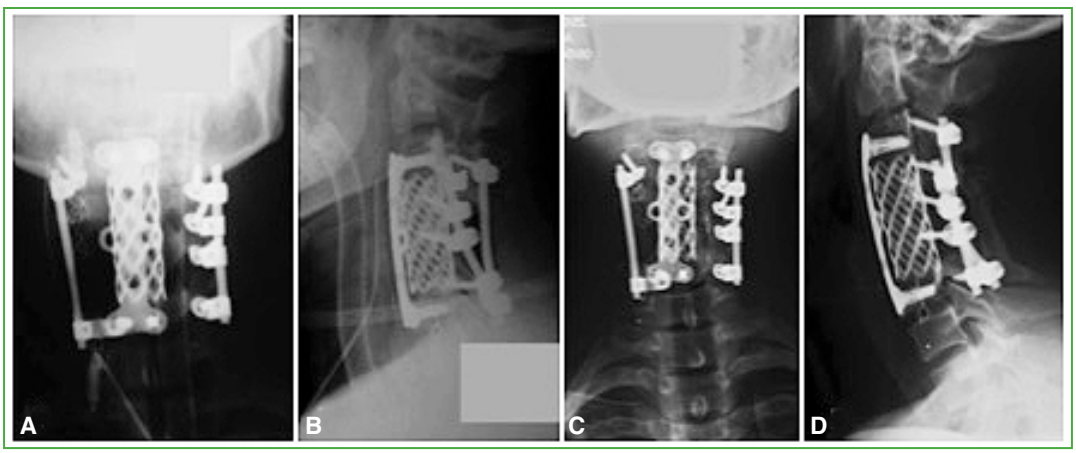

Figure 5. A and B. AP and lateral postoperative X-rays. **C and D.** X-rays at 10 years.

DISCUSSION

Although osteosarcomas are the second most common primary bone tumors, spinal involvement is rare and varies between 3% and 5% of all osteosarcomas,[8,9] and 3-14% of malignant spinal tumors. The most common symptom of osteosarcomas is pain, presenting in almost all patients, while up to 70% of them may have neurological impairments.[12] Presentation and radiological findings vary, so late diagnosis is the most common situation.[7] The main differential diagnosis is osteoblastoma. Both osteoblastomas and osteosarcomas are bone-forming neoplasms arising from the bone marrow. Osteoblastoma-like osteosarcomas are a rare variant of low-grade osteosarcomas that resembles an osteoblastoma. Radiological findings can vary from a lytic to a sclerotic bone lesion, and its borders may appear well to poorly defined, making it even more difficult to diagnose.[5] Distinguishing these two conditions is very important to define prognosis and treatment. Upon histopathological examination, osteoblastomas are generally characterized by a loose fibrovascular connective stroma that separates the osteoid trabeculae, and by their lack of permeation. In cases of osteosarcoma, the medullary spaces between the trabeculae are occupied by malignant cells. If the biopsy allows to observe the periphery of the lesion, osteoblastomas may show a well-defined border separating it from normal bone. In contrast, osteosarcomas tend to invade adjacent normal cancellous bone.

Currently, it is known that *en bloc* resection with free margins is the best surgical approach to eradicate primary tumors of the spine.[13,14] Bertoni *et al.*[7] reported 11 cases of osteoblastoma-like osteosarcomas at the Rizzoli Institute. They observed different radiological patterns—from lytic to sclerotic—and reported a high recurrence rate after performing intralesional curettage in 5 patients, compared to those undergoing wide resection.

Regarding unilateral ligation of the vertebral artery during *en bloc* resection, in a series of 15 patients, Hoshino *et al.*[15] showed that it does not cause adverse effects on the brain, cerebellum or spinal cord. However, we believe that the preoperative exam is of utmost importance, either by cranial and spinal angiography, or transient occlusion of the vertebral artery with somatosensory and motor evoked potentials.

Abramovici *et al.*[16] described an osteoblastoma-like osteosarcoma of the distal tibia in a 14-year-old boy who underwent an *en bloc* resection followed by arthrodesis after recurrence due to poor curettage. Tani *et al.*[17] also reported recurrence after curettage and bone grafting of a proximal femur, which ultimately resulted in an osteoblastoma-like osteosarcoma.

In our case, the preoperative biopsy confirmed the suspicion of a malignant tumor, so we performed an *en bloc* resection with free margins as first-line treatment.

CONCLUSIONS

Diagnosis and treatment of primary tumors of the spine require a multidisciplinary and thorough management, as there are conditions they can be mistaken with, both clinically and histopathologically. On the basis of the available literature, since osteoblastoma-like osteosarcomas are aggressive tumors, a wide resection is recommended to decrease recurrence rate and as a definitive treatment followed by chemotherapy.

Conflict of interests: Authors claim they do not have any conflict of interests.

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