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CASE REPORT

# Aggressive osteoblastoma of the cervical spine involving the canal and vertebral artery: a case report

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**Abstract**
*Objective* We present such a case of aggressive osteoblastoma of cervical spine. We describe its complicated clinical progression, hoping to shed light on the surgical strategy of this complex tumor.
*Methods* We present such a case of aggressive osteoblastoma involving the C6–7 vertebrae. A 25-year-old man was diagnosed as aggressive osteoblastoma of the cervical spine. The lesion encroached upon the radicular foramina and was located adjacent to the canal of the vertebral artery. Preoperative embolization was performed to reduce intraoperative bleeding and to prevent intraoperative injury of the vertebral artery.
*Results* A pathologic examination showed osteoblasts suggestive of osteoblastoma. At 2-year follow-up, bony union was achieved, and there was no evidence of recurrence on a CT scan.
*Conclusion* En bloc total resection for highly vascular osteoblastoma is ideal, but this case shows that piecemeal total resection following preoperative embolization is a surgical option for highly expansive osteoblastoma.

**Keywords** Aggressive osteoblastoma · Cervical spine · Involving the canal and vertebral artery

## Introduction

Osteoblastomas are relatively rare osteoid-producing benign primary bone tumors that account for 1% of all primary bone tumors and 3% of benign bone tumors [2, 12]. These tumors are rarer than osteoid osteomas, with a reported incidence of 10–25% of primary osseous spine tumors [3]. Osteoblastomas typically involve the posterior elements of the spine, but due to their larger size may extend into the anterior vertebral body and canal [14]. In 1956, Lichtenstein and Jaffe first reported a series of osteoblastomas [15, 17]. By definition, the lesion size of an osteoblastoma at diagnosis is above 2 cm [19]. In the Enneking staging system [10], stage 3 osteoblastomas are defined as tumors with fully lytic lesions eroding the cortex and invading the canal and/or surrounding soft tissues. These tumors are referred to as aggressive osteoblastomas [9]. The aggressive nature is illustrated in a study by Raskas et al., in which 83 of 149 (56.6%) osteoblastomas invaded the epidural space and required dissection of the dura mater, whereas these features occurred in none of 159 osteomas [21].

Resection is the standard treatment for osteoblastoma, but local recurrence after surgery, especially subtotal resection, occurs for approximately 50% of Enneking stage 3 spine lesions, and en bloc resection is recommended for aggressive osteoblastomas [5]. However, the cervical spine has complicated anatomic constraints, such as the dura mater of the spinal cord, spinal root, and vertebral artery, which cause difficulty in en bloc resection. Moreover, aggressive osteoblastomas are highly vascular tumors, which further complicate complete resection.

To the best of our knowledge, there has been no report in the English literature of a case of cervical osteoblastoma involving two vertebral bodies and posterior elements that caused myelopathy. Here, we present such a case of

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aggressive osteoblastoma involving the C6–7 vertebrae. The patient underwent preoperative embolization to reduce blood loss, followed by tumor resection and spinal fixation. The patient and his parents gave informed consent for submission of this case study for publication.

## Case report

A 25-year-old man presented with neck pain, gradually progressive difficulty in walking, and numbness in both hands for 6 months. An abnormal cervical lesion had been detected incidentally at a different hospital after a traffic accident, and he was referred to our hospital for further management. Preoperative examination revealed mild hyperesthesia in both hands and atrophy in the right-hand intrinsic muscles. The patient could perform the one leg standing test on each leg, but only for 2 s. He was hyporeflexive in TTR and hyperreflexive in the lower limbs without a bilateral Babinski response. His preoperative Japanese Orthopaedic Association (JOA) score was 10/17 [1]. He had no other past history.

Routine laboratory blood tests were all within the normal range. Chest X-ray was unremarkable. Plain cervical X-rays revealed a radiolucent lesion located in the pedicle and facet on the right side, which was visible in the anteroposterior view of C6–C7 (Fig. 1). Reconstructed computed tomography (CT) of the cervical spine showed an expansive and highly vascular osteolytic lesion with a thin rim of cortex involving the spinous process, lamina, pedicle, superior articular process, transverse process, and vertebral bodies of C6–7 (Fig. 2). The lesion encroached upon the radicular foramina and was located adjacent to the canal of the vertebral artery. Secondary spinal canal narrowing was also noted.

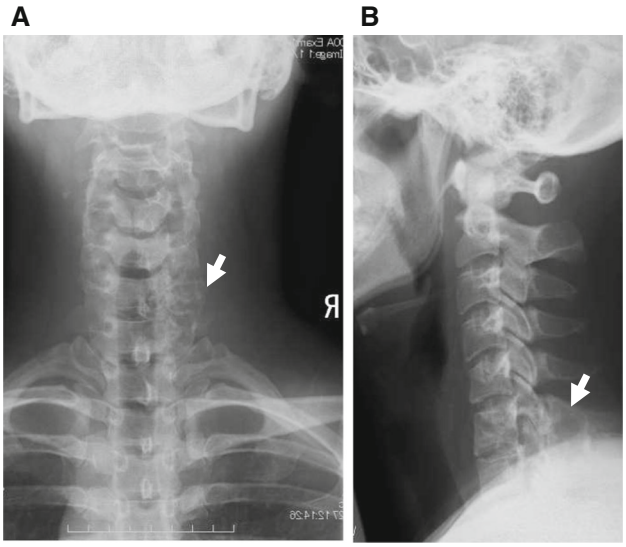


**Fig. 1** **a**, **b** Plain cervical X-rays revealed a radiolucent lesion located in the pedicle and facet on the right side that was visible in the anteroposterior view of C6–C7 (*arrow*)

Sagittal cervical magnetic resonance imaging (MRI) showed an expansile, well-demarcated, inhomogeneous (iso in T1-weighted images and hypo- and hyperintense in T2-weighted images) mass that extended into the canal at the C4-T1 levels (Fig. 3a, b). Gadolinium-enhanced images showed destruction of mainly the right side in the C6 and C7 vertebra, pedicle, lamina, spinous process, and facet joint (Fig. 3c). Axial MRI displayed encircling of the right vertebral artery (Fig. 3d). A pre-surgical open-biopsy under general anesthesia was performed, and pathological evaluation was osteoblastoma without malignant features. The patient was treated with vertebral artery embolization with platinum coils introduced via an endovascular technique using fluoroscopic guidance before surgery to reduce intraoperative blood loss (Fig. 4).

Two-stage surgery was planned. During the first operation, a midline incision was performed using Mayfield tongs with the patient in a prone position. The paravertebral musculature was gently dissected from the posterior tumor mass and the capsule was left intact. The spinous processes and laminae of C4 and T2, as well as the lateral masses, were exposed, thus clearly identifying the border of the tumor mass. The tumor mass was removed in a piecemeal fashion, displaying the dura and the exiting right C7 nerve root, which was completely exposed through a foraminotomy. Through further tumor mass removal, the embolized vertebral artery was exposed circumferentially. Finally, tumor tissue was removed from the right dorsolateral end of the C6–7 vertebral bodies. Posterior instrumentation of C4-T2 was performed with left C4 and C5 lateral mass screws, right C4, 5, left C7 (short screw for anterior approach), and bilateral T1 and T2 pedicle screws. An O-arm navigation system was used during insertion of screws. The remaining lateral masses were shaved using a high-speed burr, and cancellous bone from the iliac crest was grafted. A crosslink was added to the instrumentation to enhance stability in axial rotation. A deep and subcutaneous drain was placed and wound closure was performed. The total blood loss for the procedure was 1800 mL and the patient did not require blood transfusion.

After 10 days, total resection of the residual bony mass was performed through an anterior approach between the sternocleidomastoid muscle and carotid sheath. The remaining tumor was removed with the embolized vertebral artery. The circumferential margin of normal bony tissue was also removed with a high-speed burr. The C6–C7 bodies were reconstructed with the iliac crest (Fig. 5a, b). The total blood loss for the second procedure was 100 mL and the patient did not require transfusion.

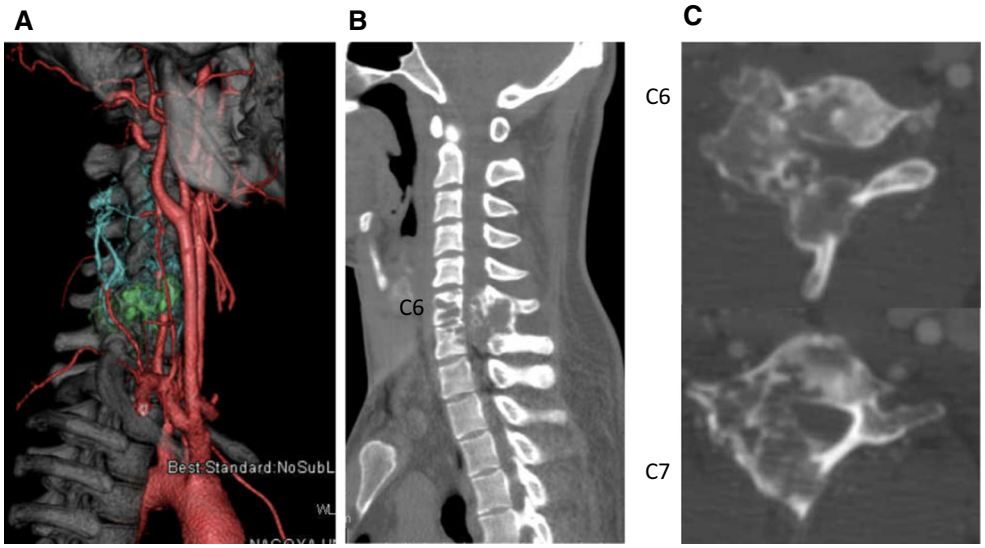


**Fig. 2** **a–c** Reconstructed computed tomography (CT) of the cervical spine showed an expansive osteolytic lesion with a thin rim of cortex and high vascularity. The lesion involved the spinous process, lamina, pedicle, superior articular process, transverse process, and vertebral bodies of C6–7

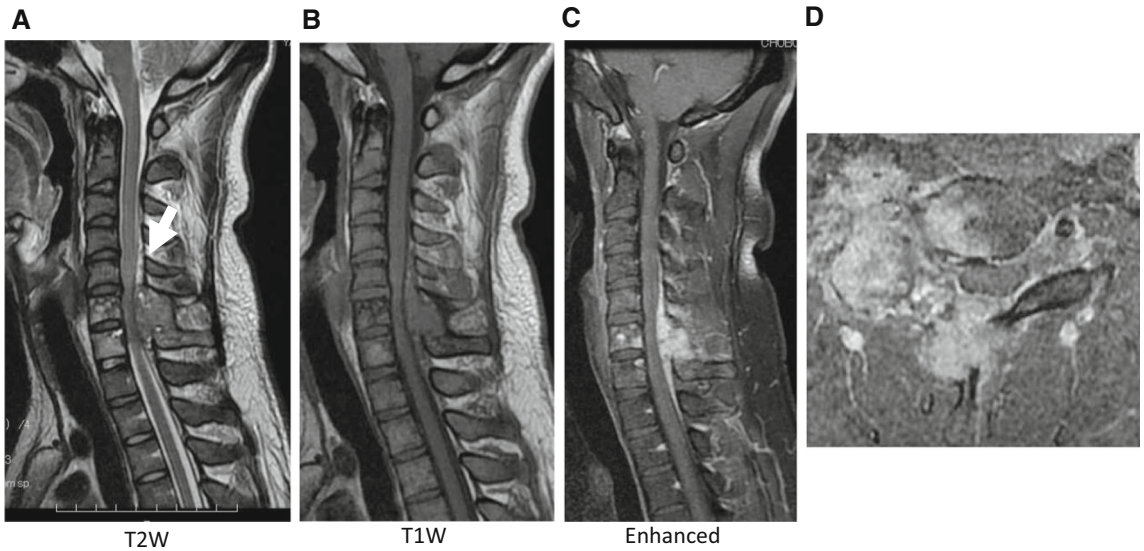


**Fig. 3** **a**, **b** Sagittal cervical magnetic resonance imaging (MRI) showed an expansile, well-demarcated, inhomogeneous (iso in T1-weighted images and hypo- and hyperintense in T2-weighted images) mass that extended into the canal (*arrow*) at the C4-T1 levels. **c** Gadolinium-enhanced MRI depicted destruction of mainly the right side in the C6 and C7 vertebra, pedicle, lamina, spinous process, and facet joint. **d** Axial MRI displayed encircling of the right vertebral artery

## Pathologic examination

A pathologic examination showed osteoblasts suggestive of osteoblastoma. None of the tumors had significant mitotic activity and the MIB-1 indices were all <5%.

## Follow-up

The postoperative course was uneventful. The patient was immobilized with a cervical collar for 3 months. Neck pain disappeared soon after the operation. Motor weakness and

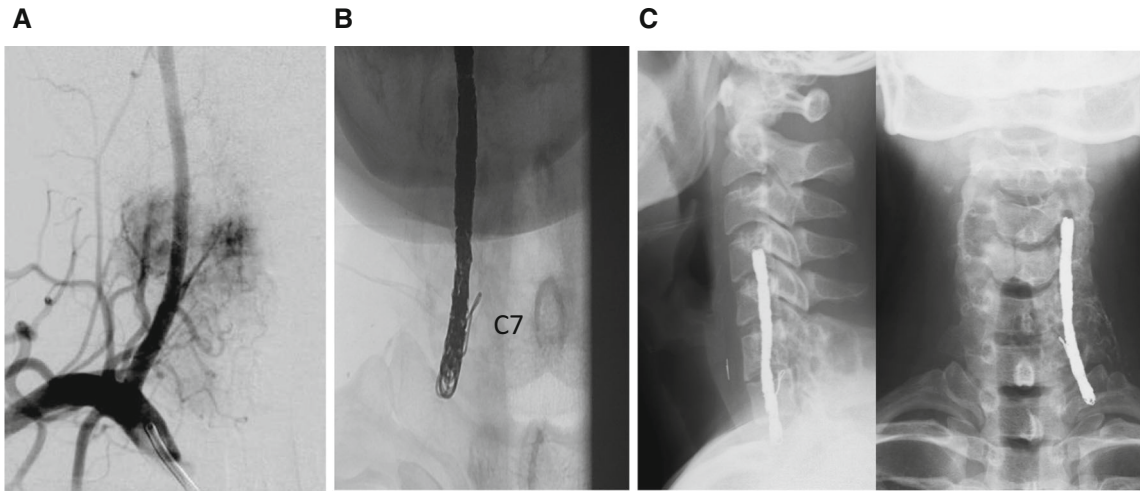


**Fig. 4** **a–c** Vertebral artery embolization with platinum coils introduced via an endovascular technique using fluoroscopic guidance was performed before surgery to reduce intraoperative blood loss

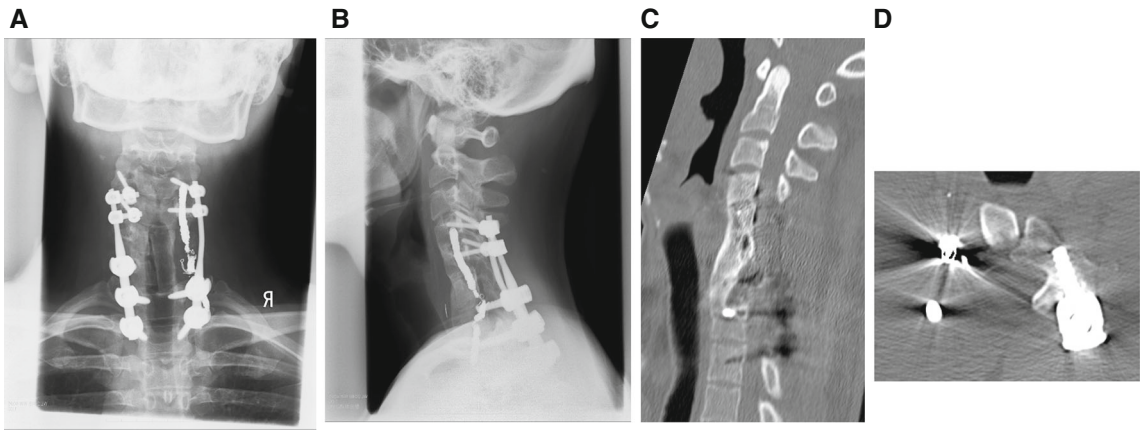


**Fig. 5** **a–d** At 2-year follow-up, CT showed bony union and no evidence of recurrence

sensory disturbance improved within 3 months, except for the C7 dermatome and atrophy of right-hand intrinsics. At 2-year follow-up, the patient presented with full neurologic recovery, bony union was achieved, and there was no evidence of recurrence on a CT scan (Fig. 5). The JOA score improved to 15/17.

## Discussion

We have presented a case of aggressive osteoblastoma with cervical myelopathy caused by tumor canal involvement. Osteoblastoma is histologically similar to osteoid osteoma, but occurs in slightly older patients, has a greater propensity for the spine, and is more biologically aggressive, with a tendency to form a less sclerotic but more expansive mass [7, 29]. Expansively growing cervical spine osteoblastomas are rare, but can cause severe neurological damage as a result of their anatomical relationship to nerve structures [25].

The rate of malignant transformation is 12–25% [5, 18, 23, 27]. Rapid growth over months has been described for aggressive osteoblastomas [20, 28]. Surgery is the treatment of choice for relief of symptoms, stopping the aggressive behavior that may destroy neighboring vital neurovascular structures, and preventing malignant transformation [13]. The surgical approach for resection of spinal tumors is selected based on a host of factors that include the type of tumor and its location within the spinal column, the presence or absence of neural compression, the portion of spinal cord involved, and the stability of the spine [4, 27, 30].

In a review of 40 Enneking stage 3, osteoblastomas treated over 26 years from 1984 to 2010. Boriani et al.

found incidences of recurrence of 0% (0/10) after wide/-marginal en bloc resection and 7% (2/28) after intralesional excision with or without radiotherapy [5]. However, good results can be obtained with intralesional excision [6, 16, 22] and anatomic aspects of the cervical spine make en bloc resection difficult for aggressive osteoblastomas. In the current case, we performed intralesional total resection, rather than en bloc resection, because gadolinium-enhanced MRI revealed extraosseous inflammatory reactions associated with the Enneking stage 3 osteoblastoma at the C4-T1 levels, which made en bloc resection impossible. A two-stage surgical approach for resection was performed, because the tumor was an expansive osteolytic lesion.

Aggressive osteoblastomas are highly vascular tumors [26], which also make complete resection difficult. Liang et al. reported extensive intraoperative bleeding (12,000 ml) in a case in which preoperative embolization was not performed [16], and other reports have indicated the potential utility of preoperative embolization [8, 16, 24]. In the current case, preoperative embolization was performed to reduce intraoperative bleeding and to prevent intraoperative injury of the vertebral artery [11]. Moreover, to minimize the risk of microscopic tumor residues, our procedure involved exposure of the whole lesion through the normal tissue and removal of the lesion and reactive tissue by piecemeal total resection. However, adjuvant radiotherapy should be considered if recurrence in case of such intralesional excision.

## Conclusion

We have reported the case of a 25-year-old man with cervical aggressive osteoblastoma that involved two vertebral bodies and posterior elements, and resulted in myelopathy. En bloc total resection for highly vascular osteoblastoma is ideal, but this case shows that piecemeal total resection following preoperative embolization is a surgical option for highly expansive osteoblastoma.

#### Compliance with ethical standards

**Conflict of interest** None of the authors has any potential conflict of interest.

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