

## Iatrogenic vertebral artery injury during cervical spine surgery: A case report

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### Abstract

Osteoblastomas are rare benign, primary bone tumors which frequently arise in the spine. Patients often present with dull back pain sometimes associated with painful scoliosis. Neurological manifestations such as radiculopathy or myelopathy are due to mass effect on nerve roots or the spinal cord. Optimal treatment is complete surgical resection, preceded by embolisation. The preoperative radiological assessment is extremely important for identifying anatomical risk factors.

### Presentation of case

A 19 years-old-man, presenting with a history of cervico-brachial neuralgia and weakness of the right upper limb lasting from 18 months. The CT scan showed a bone tumor of the posterior arch of C5 and invading the transverse foramina. While removing the tumor, an injury of the vertebral artery occurred.

### Conclusion

Osteoblastomas have a high risk of relapse and can potentially degenerate in sarcoma. Aggressive total resection is the preferred treatment, but it is risky regarding to the proximity of the vertebral artery, so surgery must be meticulously planned. This case points the radiological, histological and therapeutic features of osteoblastomas and surgical difficulties encountered during resection.

### Keywords

Aggressive osteoblastoma; vertebral artery injury; primary spine tumor.

### Introduction

Osteoblastomas (OBL) are rare benign, primary bone tumors that affect mainly the long bones. Thirty –six percent of these tumors are observed around the spine and the vast majority arises around the posterior elements (1). In the literature, there are about 120 OBL of the spine. In 1984, Dorfman and Weiss (2) describe for the first time a subtype of OBL, they called aggressive osteoblastoma (AO) and define it as a borderline tumor with epithelioid osteoblasts, endowed with a high potential of recurrence and sarcomatous transformation. Since then, rare cases of AO have been published. Optimal treatment is complete surgical resection, preceded by embolisation. Adjuvant radiotherapy is indicated according to quality of resection and criteria of aggressivity (3). This report describes a case of a 19-year-old man operated on an OBL of the posterior C5 arch, with clinical-radiological criteria of aggressiveness. Total resection was more complicated than expected. The clinical presentation, radiological evaluation and management difficulties are presented.

The work has been reported in line with the SCARE criteria (4).

### Presentation of case

This is a 19-year-old man with no medical history, who has been complaining for 18 months of right cervico-brachial C5 neuralgia radiating from the neck to the shoulder and the right upper limb. This pain was initially triggered by the effort, then became permanent not responding neither to the rest, nor to the analgesic and anti-inflammatory treatment. Four months ago, a weakness of the right upper limb appeared, motivating the consultation.

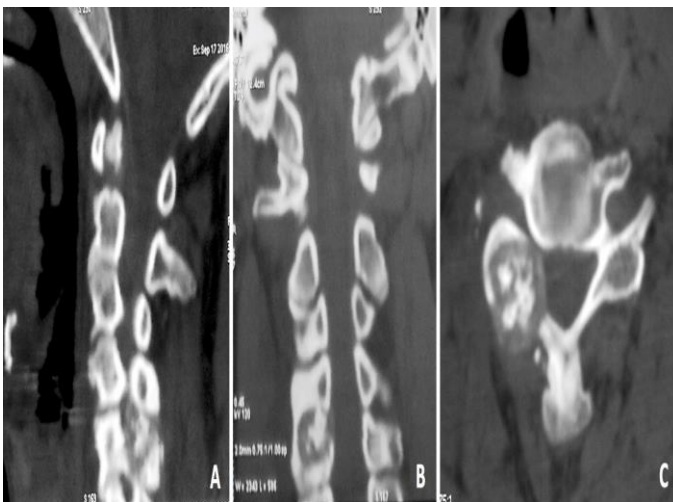
On clinical examination, stiffness of the cervical spine was noted with an antalgic attitude of right lateral inclination of the neck and elevation of the right shoulder. There was exquisite pain in the pressure of the last cervical spinous processes and a right paravertebral muscle contracture. Muscular testing was normal, but a slight amyotrophy of the right shoulder girdle was noted. The remainder of the clinical examination was normal, including no sensory disturbances or abnormalities of the osteo-tendinous reflexes.

Radiological investigations included standard radiographs of the cervical spine, face, profile, right and left (Fig. 1), medullary MRI and cervical CT scan.



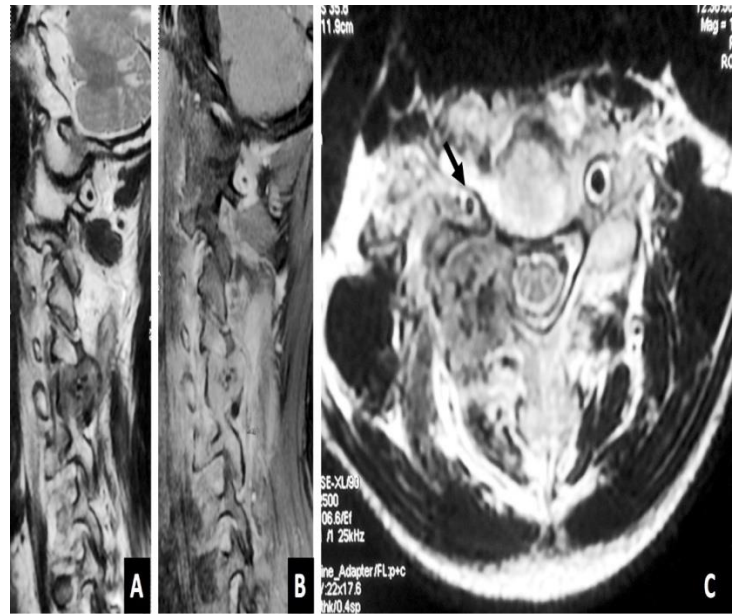
**Fig. 1 : Preoperative radiographs. (A) :** Anteroposterior view shows a right high intensity area without clear margins projecting on C4-C5 articulation. **(B) :** Lateral radiograph shows a mild osteolysis in the pedicle and lamina of C5 (Arrow). These radiographs were read as « normal » before worsening of the clinic signs motivating a CT scan.

Cervical CT scan showed an osteolytic bone tumor of the right lamina of C5, of approximately 22 mm in diameter, containing round and linear calcifications associated with hyperostosis. There is a rupture in the cortical bone. This lesion narrows considerably the C5 foramen, responsible of a C5 root compression, and invades the transverse foramen, coming into contact with the right vertebral artery (VA) (Fig. 2).



**Fig. 2: Sagittal (A), coronal (B) and axial (C) Computed Tomography images** demonstrating an expansile lytic tumor at the level of the right C5 lamina and articulation with epidural extension and foraminal narrowing. The cortical bone is ruptured by a blowing tumor containing round central microcalcifications

MRI showed an expansile tumor of C5 articulation, with ipsilateral postero-lateral epidural extension, invasion of the radicular foramina and reduction of the diameter of the VA channel. The right VA is compressed by the tumor and its lumen is narrowed. The adjacent paraspinal soft tissues were site of a diffuse inflammatory reaction. MRI showed also a diffuse hypo T1, T2 and STIR hypersignal remodeling of C4 and C5 body, which intensified after Gadolinium injection (Fig. 3).



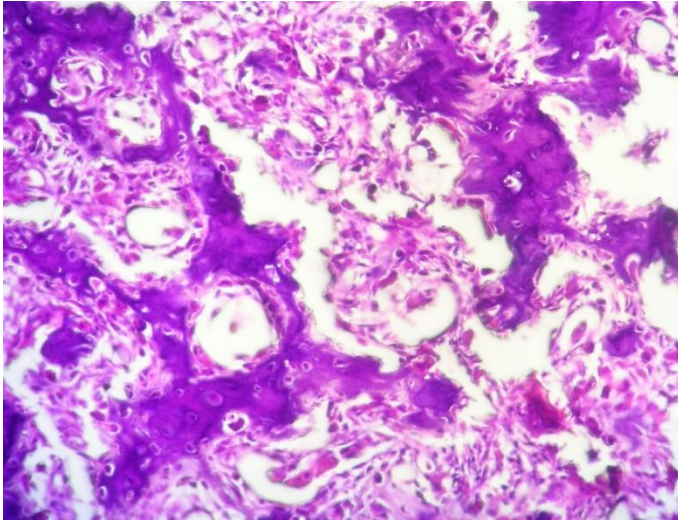
**Fig. 3: MRI: (A) sagittal T2, (B) sagittal FAT SAT Gadolinium (C) and axial FAT SAT Gadolinium images,** showing an expansile tumor of the C5 articulation, with intralésional signal void corresponding to matrix calcifications. The lesion encroaches upon the radicular foramina, reaches the canal of the vertebral artery (Arrow). The latter is significantly reduced in diameter. There is an epidural extension without medullar compression. Hyper T2 intensity of the adjacent vertebrae and surrounding paraspinal soft tissues is due to diffuse reactive inflammatory response

The patient underwent a scan-guided biopsy of the lesion, but the anatomopathological result was inconclusive (non-specific inflammatory tissue).

Preoperative embolization was recommended, but not performed due to lack of technical platform.

After anaesthesia, the patient was placed carefully in a "concorde" position, with a Mayfield 3-point head holder. A posterior cervical spine approach was performed exposing the posterior arches of C3 to C6.

We found that the tumor has obviously increased in size and spread, compared to MRI dating from two months before surgery. Indeed, this tumor is currently about 3.5 cm, occupies the C4-C5 articulation, extends through the laminae of C4 and C5 to the spinous processes. The blown articular mass compresses the C5 root and the right lateral side of the spinal cord. This tumor is red hemorrhagic, friable. Total tumoral excision was carried by intratumoral morcelling and drilling, removing the C4-C5 articular mass. A right C4 and C5 hemi-laminectomy with the spinous processes was also necessary. Section slices were normal. At the end, wanting to lift up a fragment of cortical articular bone using a dissector, a very abundant jet of arterial bleeding suddenly appeared, originating from a laceration of the right VA. The injury was clogged by compression, aponeurosis packing and biological glue. The hemodynamic state remained stable during the three hours of intervention despite the abundant bleeding (11) and the patient did not need blood transfusion. The operative follow-up was simple, the patient was extubated on the operating table, immediate disappearance of cervico-brachial neuralgia, and no neurological deficit had to be deplored. He left the hospital on day 3 postoperative, with recommendation to wear a type C4 minerve permanently during the first month. The histological examination of the surgical specimen, concluded with an osteoblastoma (Fig. 4). Re-reading of the slides in search of histological signs of aggressiveness has been unsuccessful. A cervical CT Scan (Fig. 5) and an MRI were performed at 4 months showing no tumor residue. A bone scintigraphy was made at six months and showed no metastasis. Standard X-rays for assessment of cervical spine stability are regularly done. After one year of follow up the patient remains asymptomatic.



**Fig. 4: Histology results.** Sclerotic bone trabeculae with marginal osteoblastic activity (Photomicrograph : H&E, original magnification x 40).



**Fig. 5: Postoperative CT Scan:** (A) axial and (B) sagittal images: No evidence of residue or recurrence.

## Discussion

Osteoblastomas are benign primary bone tumors counting for 1% of all bone tumors, and close to 40% of them are localized in the spine (1,5). Two types of osteoblastomas have been described in the literature: conventional osteoblastomas (CO) and aggressive osteoblastomas (AO).

Whereas CO was presented clinically with nocturnal local pain, AO was more likely to cause spinal deformity and neurologic disorders than CO, because AO had a significantly larger scope of tumor involvement and size (3). Plain radiography is the most common radiological choice and is thought to have great value for diagnosing spinal OBL. However, normal images were found in 50% of the patients with CO and 25% of cases with AO, which might cause a high number of misdiagnoses (3). The diagnosis is usually made by computed tomography (CT). Osteoblastoma appears as an inhomogeneous lytic and sclerotic mass (stages 1 and 2 according to the Enneking staging system for benign lesions) (6). As it was the case with our patient, some osteoblastomas appear more aggressive and can demonstrate larger osteolytic components (stage 3 lesions). Magnetic resonance imaging (MRI) complements CT imaging and gives information on possible spinal cord, nerve root or vertebral artery (VA) compression.

Histologically, the invasion of cortical bone, the larger epithelioid osteoblasts and the increased synthesis of alkaline phosphatase permit to distinguish AO from CO (5). Total surgical resection is recognized to be the most effective treatment for OBL in the spine by reducing relapse rates (7, 8).

OBLs are richly vascularized tumors, causing significant intraoperative bleeding, and especially when it is an AO. In fact, this blood loss has been estimated at 2.3 l on average for AO and 1.1 l for CO (3). This difference is due to the larger size of AO which implies a longer operating time. In addition, in some cases, the tumor erodes the VA canal, and a wound in the VA can occur, as in our patient's. A similar case has been found in the literature (9), where complete excision of the tumor could only be achieved at the cost of a wound in the VA. In both cases, the bleeding was controlled by compression by multilayer packing and biological glue, then replaced the paravertebral muscles to ensure against pressure and prevent rebleeding.

In both two cases, the angiography associated with tumor embolization was not made. If it had been done, it would have significantly reduced intraoperative tumor bleeding (10), and studied the AV surrogacy network, which would have allowed, in the event of a vertebral artery injury (VAI), to clip the VA without wondering about ischemic consequences (cortical blindness...) (11).

In general, the resection of an OBL of the cervical spine poses a significant morbidity problem of the VA, since the OBL is often in contact with the transverse foramen, and may even include the VA. In these cases, it would be wiser to perform an intratumoral resection instead of the ideally recommended marginal resection, to avoid unnecessary morbidity (12). The use of navigation and / or intraoperative scanning would reduce the risk of AV wounds during resection. A displacement of the VA before the removal of the OBL is proposed whenever possible (7). Radiotherapy and chemotherapy, either together or individually, have been used for patients with unresectable lesions or in cases of recurrent disease (13, 14).

In the literature, VAI during cervical spine surgery are extremely rare. A recent study demonstrates an incidence of 0.08% of VAI during cervical spine surgery (15). Anterior cervical spine surgery is associated with the highest rates of VAI. Surgical team have to identify anatomical risk factors of VAIs injuries (anomalous vertebral artery, tortuous vessel course, and medial migration of the artery) using advanced imaging techniques such as angiography, reformatted 3D CT or MRI images.... VAI outcomes are variable. The majority of the patients have favorable outcome. Despite this, VAI still be a serious complication which can lead to perioperative death and post-operative neurologic sequelae. The surgical team must be prepared to face this complication in case it occurs.

## Conclusion

Aggressive osteoblastomas have a high risk of relapse and can potentially degenerate in sarcoma (16). Aggressive total resection is the preferred treatment, but it is risky regarding to the proximity of the vertebral artery. Detecting clinic-radiological characteristics of aggressivity helps planning surgery in a way to avoid per operative bleeding, and achieve complete resection.

## List of abbreviations

OBL : osteoblastoma

CO : conventional osteoblastoma

AO : aggressive osteoblastoma

VA : vertebral artery

CT : computed tomography

MRI : Magnetic resonance imaging

VAI : vertebral artery injury

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