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Case Report

Aggressive osteoblastoma of the cervical spine and resultant complication due to swollen oxidized regenerated cellulose: A case report

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ABSTRACT

Background: Osteoblastomas, although rare, are benign primary bone tumors, with cervical spine involvement being exceptionally uncommon. Late diagnosis, especially in aggressive cases, can lead to surgical challenges. Oxidized regenerated cellulose (ORC) used for hemostasis may result in complications if left in the surgical field.

Case Description: An 8-year-old female presented with six months of intractable neck pain accompanied by swelling, hindering proximal right upper extremity evaluation. Motor strength was intact distally, with normal reflexes and no hypoesthesia. Imaging revealed a C4–5 facet joint lesion necessitating surgery. Intraoperative hemorrhage prompted ORC application, which led to postoperative arm pain and C5–6 radiculopathy. Subsequent surgery alleviated these symptoms.

Conclusion: Osteoblastomas, despite their benign classification, may exhibit aggressive characteristics, warranting *en-bloc* resection. Cervical spine osteoblastomas, due to their vascular nature and proximity to vital structures, complicate surgical interventions. ORC, a commonly used hemostatic agent, may induce compression complications, and early intervention is critical for patient recovery. This case underscores the intricacies of managing aggressive osteoblastomas in the cervical spine and highlights potential ORC-related complications. Surgeons must exercise caution when using ORC and consider postoperative risks. Prompt intervention and meticulous planning are paramount for favorable outcomes in such cases.

Keywords: Cervical spine osteoblastoma, Complication, Swollen oxidized regenerated cellulose

INTRODUCTION

Osteoblastomas are rare and benign primary bone tumors that account for 1% of all primary bone tumors. About 40% of them occur in the spine, predominantly involving the posterior elements.[2,6,18] Osteoblastoma cases of the cervical spine are rarely reported.[1,5,9,10,12,15] Due to its low incidence and nonspecific symptoms during the early course, late diagnosis is highly likely.[9,12,15] If the lesion has progressed to aggressive osteoblastoma (Enneking Stage 3), *en bloc* resection is recommended for treatment.[1,2] The proximity of the spinal cord, nerve roots, and vertebral artery at the cervical spine region and the highly vascular nature of aggressive osteoblastoma make total resection challenging and increase the risk of complications.[1] Oxidized regenerated cellulose (ORC) (Surgicel®) is widely used as a hemostatic agent in neurosurgery.

Although complete removal of ORC after hemostasis is recommended, leaving a certain amount in place is often preferred to prevent postoperative hematoma.[11,17] However, leaving the ORC at the surgical site can also potentially lead to complications.[3,4,7,8,11] This case report presents an 8-year-old patient with aggressive osteoblastoma at the C4–5 facet joint and complication of paresis due to swollen ORC.

CASE

An 8-year-old female patient presented with a 6-month history of progressively worsening neck pain and tenderness, unresponsive to anti-inflammatory medication. Physical examination revealed swelling in the right posterior neck and increased pain during shoulder movements and palpation. Due to pain, a comprehensive assessment of proximal movements of the right upper extremity could not be performed, but normal motor strength was observed distally. Deep tendon reflexes were bilaterally normal, and no hypoesthesia was detected. A computed tomography (CT) scan revealed a 21 mm lesion at the level of the C4–5 right pedicle and articular facet, extending to the lamina and showing minimal protrusion into the spinal canal. CT angiography imaging confirmed that the lesion was not associated with the vertebral artery. Gadolinium-enhanced magnetic resonance imaging (MRI) demonstrated a contrast-enhancing mass with ossification signals in the central and outer matrix [Figure 1]. In addition, there was widespread edema in the paraspinal muscles between the C3 and seven levels. A posterior approach was selected, exposing the lamina and facet complexes between the C4 and six levels. The deformed area in the C5 lamina caused by the tumor was identified. Tumor excision was initiated, involving the surrounding normal bone tissue using a piezoelectric osteotome.

The tumor was removed in fragments, with a significant hemorrhage encountered during the excision of the final portion adjacent to the vertebral artery. We were aware that bleeding might be encountered during excision due to the high vascularity of the tumor. However, the bleeding flow rate exceeded our estimation (approximately 250 cc until hemostasis), which intraoperatively prevented us from distinguishing whether the cause of bleeding was of vertebral artery origin. Hemostasis was promptly achieved through the application of ORC compression. No additional hemostatic agent was applied. To mitigate the risk of postoperative hemorrhage recurrence, a portion of the ORC was intentionally retained in the surgical area. Following surgery, the patient experienced severe arm pain and exhibited grade 2/5 muscle strength in arm abduction and elbow flexion, indicative of C5–6 radiculopathy. Imaging studies revealed no damage to the vertebral artery but identified a mass within the excised tumor bed compressing the C5 nerve root, displaying hypointensity on T2-weighted MRIs [Figure 2]. It was suspected that postoperative hematoma or ORC swelling might have contributed to this compression.

Consequently, the patient underwent a second surgery for exploration and decompression the following day. During the second operation, no hematoma was observed, but the previously retained ORC had expanded due to swelling. Most of the ORC was removed to alleviate compression on the dura and nerve root, with a minimal amount left in the area where hemorrhage had occurred. Subsequently, the patient's severe arm pain subsided, and muscle strength improved to an early-stage grade of 3/5 for the deltoid, biceps, and brachioradialis. Following eight months of physiotherapy, the patient regained a muscle strength grade of 5/5. Pathological examination of the surgical specimen confirmed the presence of osteoblastoma [Figure 3]. In the 12-month follow-up, the patient demonstrated complete neurological recovery, and MRI scans revealed no evidence of recurrence.

DISCUSSION

Although classified as a benign tumor, osteoblastoma shows high vascularity and can exhibit locally aggressive growth. Typically, it involves the posterior elements of the spine.[12,15] While cervical spine involvement is rare, the proximity

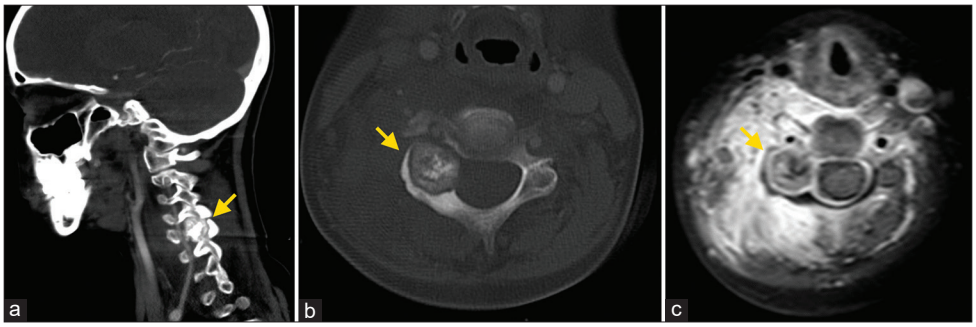

Figure 1: (a) Sagittal angio-CT scan indicate the presence of a tumor (arrow) at the level of the C4-5 right pedicle and articular facet. (b) The inhomogeneous matrix ossification is presented in the tumor (arrow). (c) Gadolinium-enhanced axial MRI depicted a well-demarcated tumor (arrow) and edema of the paravertebral soft tissue.

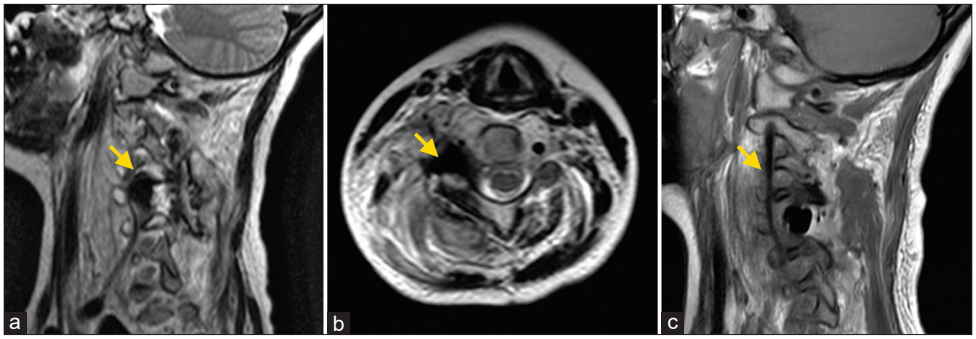

Figure 2: (a) Post-operative sagittal T2-weighted MRI depicting a hypointense mass lesion (arrow). (b) Axial T2-weighted MRI scan showing compression of the right C5 nerve root trace by the hypointense mass (arrow). (c) Post-operative sagittal T1-weighted MRI showing vertebral artery (arrow) trace.

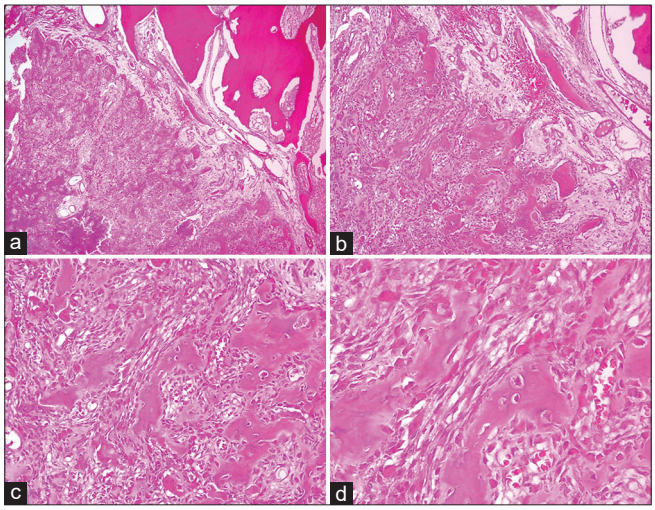

Figure 3: (a) Expansile tumor surrounded by a sclerotic rim, showing a spectrum of bone maturation changes (H&E, ×40). (b) Neoplasia with diffusely distributed osteoclast-type multinucleated giant cells with extravasated erythrocytes (H&E, ×100). (c) Bone trabeculae are covered by a single layer of osteoblasts (H&E, ×200). (d) Benign bone tumor with degenerative cytological atypia characterized by cells with large degenerated nuclei and smudged chromatin (H&E, ×400).

of the vertebral artery and the spinal cord makes total resection challenging.[1,5,15] In spinal osteoblastoma series, recurrence rates are between 9.7% and 15%, with incomplete resection being the primary cause.[9,12] The rate of malignant transformation to osteosarcoma is around 12–25%.[1,2,12,15,16] *En bloc* resection is recommended for treatment, particularly in aggressive Enneking Stage 3 tumors with invasion exceeding the bone cortex.[1,2,9,12,14,18] The most common presentation is persistent neck pain, local tenderness, and torticollis, which do not improve with nonsteroidal anti-inflammatory drugs.[5,14,18] While diagnosis can be easily made using CT and MRI scans, the delayed diagnosis rate is high due to the low incidence and nonspecific symptoms.[5,9,14,15] The prevention of total resection due to intraoperative massive bleeding can be encountered in hypervascular tumors like osteoblastoma. Therefore, some authors recommend preoperative embolization as a potential approach.[12,15] This case report presents a rare case of osteoblastoma where intraoperative heavy bleeding occurred, highlighting the complication caused by ORC used for hemostasis.

ORC is often preferred in neurosurgery to achieve hemostasis. Although it is a biologically absorbable material, it is recommended to remove ORC altogether after achieving hemostasis.[11,17] However, in many cases, it is preferred to leave a minimal amount in the surgical area. Especially in cases where intraoperative massive bleeding is encountered, the amount of ORC left in the bleeding site can be relatively high to support its tamponade effect. When ORC comes into contact with blood, it can swell and acquire a gel-like consistency, potentially causing mass effect and neural compression.[3,4,11] In the patient presenting with postoperative neurological deficit, the MRI findings resemble those of an acute hematoma. On T2-weighted images, an acute hematoma is also seen as hypointense, but the hypointensity is not as deep as that of ORC. However, a thin peripheral hyperintensity can be seen on T2-weighted images, which is believed to be postoperative fluid surrounding the ORC.[4,13] When a mass caused by swollen ORC leading to neural compression is detected, the surgical removal of the mass is recommended.[4,7,8] Early intervention to achieve decompression in neural tissue is essential for potential recovery from induced deficits.

CONCLUSION

We presented a case of an 8-year-old female patient with aggressive osteoblastoma involving the posterior elements of the cervical vertebra. These benign tumors, which have relatively high rates of malignant transformation and recurrence, ideally require *en bloc* total resection. However, due to their highly vascular nature, surgery can be challenging. Although it is a reliable hemostatic agent, it should not be preferred to leave ORC in the vicinity of nerve roots and the

spinal canal. When planning to leave the ORC in the surgical area, keeping its presence to a minimum is crucial. Otherwise, it may lead to unexpected complications through mass effect, necessitating a second surgical intervention.

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Ethical approval

The Institutional Review Board approval is not required.

Declaration of patient consent

Patient's consent not required as patient's identity is not disclosed or compromised.

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Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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