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Case Report

Giant cell tumor of the cervical spine: A very uncommon cause for cervical spine compression ☆

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ABSTRACT

Giant cell tumors (GCTs) are rare neoplasms, primarily found in long bones, typically affecting the epiphysis of the distal femur, proximal tibia, and distal radius. However, their occurrence in the cervical spine is exceedingly rare. Here, we present a case report of a 21-year-old female patient who presented with progressive neck pain, radiating numbness, and right hemiparesis. Radiographic imaging revealed a lytic lesion in the C3 vertebral body, further characterized by magnetic resonance imaging (MRI) and computed tomography (CT) scans. The patient underwent surgery for stabilization of the cervico-occipital hinge, decompression, and biopsy. Histopathological examination confirmed the diagnosis of a giant cell tumor. Postoperatively, the patient showed improvement in motor impairment, cervical pain, and numbness. She was proposed for adjuvant treatment based on Denosumab. However, she returned 1 month after surgery with worsened motor deficit, developing tetraparesis. Control MRI revealed a tumor flare-up. The decision was made not to reoperate on the patient and to accelerate the administration of Denosumab. Meanwhile, she experienced a pulmonary embolism leading to her demise. This case underscores the importance of considering giant cell tumors in the differential diagnosis of cervical spine lesions and emphasizes the successful and prompt management through a multidisciplinary approach involving surgical intervention and adjuvant therapy.

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Introduction

Giant cell tumors (GCTs) are primary bone tumors characterized by their benign nature but locally aggressive behavior [1]. These tumors typically arise in the metaphysis of long bones, with spinal involvement occurring in a range between 1.4% and 9.4% [2]. While GCTs are uncommon above the sacrum, their occurrence in the cervical vertebrae is exceptionally rare [2,3], constituting only 2% to 3% of all spinal tumors [1,4]. When GCTs develop within the spinal canal, they can cause neurological deficits by compressing the spinal cord [5]. Treatment typically involves wide surgical en bloc resection, although this may not always be feasible in spinal cord lesions due to the risk of vascular or neural injury [6], a risk that is amplified when the lesion is located in the cervical spine [2,4]. Adjuvant therapies such as radiotherapy and chemotherapy with Denosumab are often employed, but despite these treatments, local recurrences and distant metastases remain common [7].

Case report

We present the case of a 21-year-old female patient with no previous medical history, who presented with progressive neck pain, numbness in the right upper limb, heaviness in the right hemibody over the past 2 months. The patient denied any sphincter dysfunction and had not experienced recent trauma. Physical examination revealed right hemiparesis, predominantly affecting the right upper limb. Cervical spine X-ray (Fig. 1) demonstrated an osteolytic lesion in the vertebral body of C3. Cervical spine CT scan (Fig. 2) revealed complete destruction of the C3 vertebral body with extension into the odontoid bone. MRI of the spinal cord (Fig. 3) showed a mass extending anteriorly from the C2 and C3 vertebrae, predominantly on the right side, extending towards the spinal canal, thus causing significant compression of the cervical cord. The right vertebral artery was entirely encased by the tumor. Surgical intervention was deemed necessary. Considering the anterior location of the lesion relative to the cervical cord and its encasement of the vertebral artery, a decision was made to perform a C2 and C3 laminectomy to decompress the spinal cord and biopsy the tumor. Due to infiltration of both vertebral bodies adding to the planned laminectomy, cervicooccipital stabilization was also performed. Peroperatively, the tumor appeared fleshy and highly hemorrhagic, with no discernible cleavage plane from surrounding structures. The postoperative course was uneventful, and the patient demonstrated improvement in sensory and motor deficits. Follow-up cervical X-ray confirmed proper positioning of the cervicooccipital implants (Fig. 4), and the patient was discharged on the third postoperative day. Pathological examination (Fig. 5) concluded to a giant cell tumor. Subsequently, the patient was referred to the oncology department for Denosumab therapy. Four weeks postsurgery, the patient returned with new-onset walking difficulties and limb heaviness, occurring just 3 days before readmission. Physical examination revealed quadriplegia. There were no signs of infection at the surgical site, and laboratory tests were unremarkable. A new MRI (Fig. 6) revealed tumor flare-up, extending downwards to the level of C4. The decision was made against further surgery, and efforts were made to expedite access to Denosumab therapy. However, the patient developed sudden respiratory distress requiring intubation and mechanical ventilation. Subsequent CT angiography revealed a massive pulmonary embolism, and the patient passed away the following day.

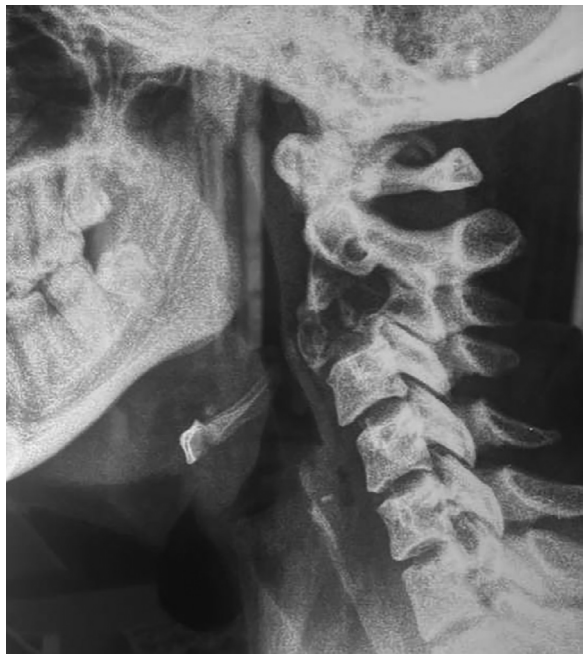

Fig. 1 – Profile radiograph of the cervical spine showing an osteolysis of the vertebral body of C3 and the lower part of C2.

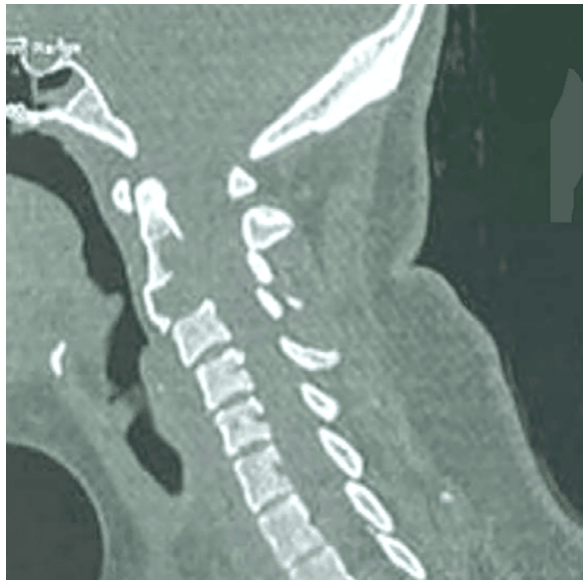

Fig. 2 – Sagittal section of a cervical spine CT scan showing an osteolysis of the vertebral body of C3 and the lower part of C2.

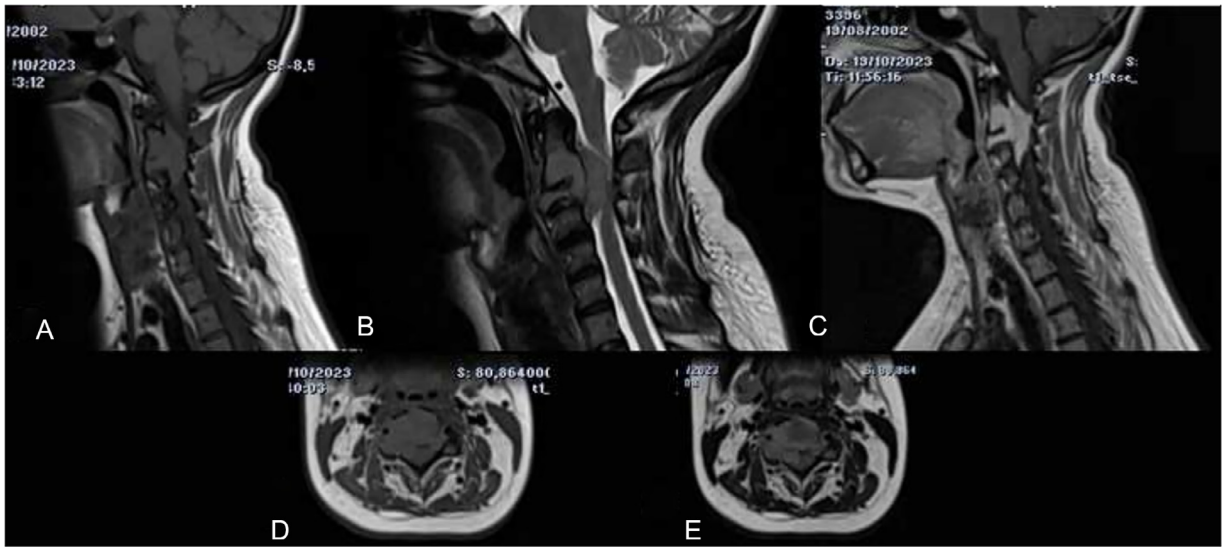

Fig. 3 – Sagittal (A-C) and axial (D, E) sections of the cervical spine on T1-WI (A, D), T2- WI (B) and T1-WI with contrast injection (C) showing a tumor of the body of C3 and the lower part of C2 extending into the spinal canal and responsible for a spinal cord compression.

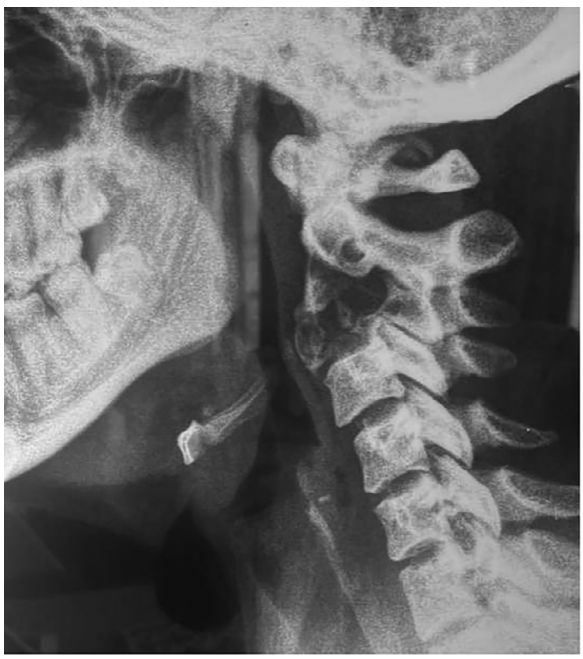

Fig. 4 – Postoperative profile radiograph of the cervical spine showing the presence of the osteosynthesis material.

Discussion

GCTs are rare tumors, and are yet more seldom and are quite rare in the cervical spine. Diagnosis has been made easier thanks to modern imaging techniques, but preoperative diagnosis still remains difficult mainly when the tumor is located in uncommon areas [2,3]. Therapeutic approach is mainly based on surgical resection, which aim is to remove the tumor and prevent its recurrence while avoiding neurological structure damage and spinal integrity deterioration [8–10]. Gross surgical resection is recommended to minimize the risk of local recurrences, but in cases like ours, resection is made difficult by the anterior location of the tumor, the encasement of large vessels, and excessive bleeding [11,12]. Anterior approach may be proposed for a wider resection and decompression, but in cases where the tumor is located on the upper cervical spine, this corridor becomes very challenging [1,6,11]. In order to improve surgical outcome for big lesions, some authors recommended preoperative embolization [8]. As quality of surgical resection frequently remains insufficient, adjuvant treatment is always proposed. Radiotherapy remains an interesting option to decrease postoperative recurrences. However, spinal irradiation remains harmful as it may be source for myelopathy, sarcomatous degenerescence, added to an important post radiation fibrosis making reinterventions highly risky [10,13]. Therefore, RT should be reserved for recurring lesions [4]. GCTs are also source for instability, a risk which is made higher by surgical bony sacrifice. It is recommended to perform multistaged resection through anterior and posterior approach with fusion, mainly when facing big tumors [14,15]. Treatment with denosumab, a receptor activator for nuclear factor kB ligand inhibitor, is considered for patients with unresectable GCTs, and to those where major nervous lesions are expected [4,7,16]. This treatment is responsible for a solidification and a reduction of the tumor [17]. Denosumab has been recommended prior to surgery [7,17], but in our case, we had no preoperative proof for the pathologic nature of the lesion. But in the specific case of cervical spine localizations, some authors reported the possibility of a short term local control using Denosumab [18]. But the long-term prognosis still remains imprecise [14,16,18]. Overall outcome is still difficult to predict as the recurrence rate vary between 11% and 50%, even following an optimal treatment [18,19]. Local recurrences are mainly encountered in the first 3 years [5,6]. Recurrences for

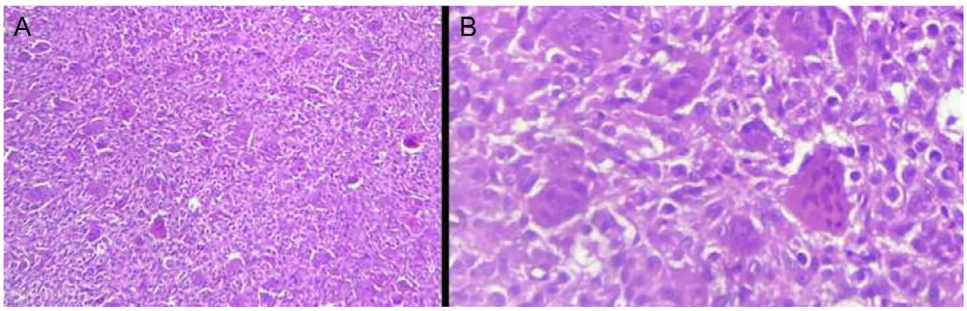

Fig. 5 – Anatomopathological sections at medium (A) and high (B) magnification showing a dense proliferation composed of multinucleated giant cells resembling osteoclasts, lacking cytonuclear atypia. Additionally, there is a sparse population of round, occasionally fusiform, mononuclear cells, few in number, and lacking cytonuclear atypia. Their nuclei are round with fine chromatin.

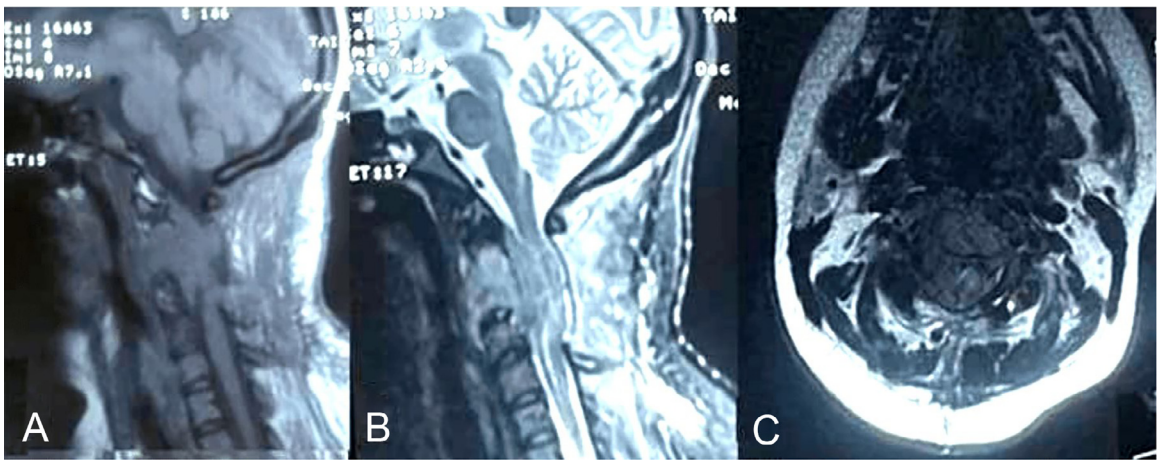

Fig. 6 – Sagittal (A, B) and axial (C) sections of a spinal cord MRI on T1-WI (A), T2-WI (B) and T1-WI with contrast injection (C) showing a tumor flare up.

spinal GCT are still difficult to define, as only a few small series related to this disorder have been reported [3,5]. But we have not been able to find case where the lesion flared up following surgical manipulation. It doesn't seem that there was any delay in giving the treatment in our case, as Denosumab is administered every 28 days with loading doses on days 8 and 15 in the first month of therapy [17]. Based on our experience, early establishment for Denosumab may prevent negative outcomes. Nevertheless, we are not able to get conclusions from this case, and wider studies may bring more concrete responses in order to clearly define the delays of beginning Denosumab therapy, especially since the use of radiotherapy in the postoperative period remains controversial.

Conclusions

The presented case underscores the challenges in managing giant cell tumors (GCTs) of the cervical spine due to their rare occurrence and potential for aggressive behavior. This case highlights the critical importance of promptly initiating adjuvant treatment following surgery for patients with GCTs, especially those involving the cervical spine, as delays can quickly lead to severe functional impairments, exacerbating the prognosis. Further research is warranted to explore alternative treatment strategies and improve outcomes for patients with this rare and challenging pathology. Additionally, efforts should be made to enhance access to advanced therapies such as Denosumab in developing countries, and to develop effective strategies for managing potential complications associated with tumor recurrence.

Patient consent

A written consent has been obtained from the patient regarding this publication.

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