

## CASE NOTE

## Seven-year survival after intralesional resection and adjuvant radiotherapy for a giant-cell tumour of the sixth cervical vertebra

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**G**iant-cell tumours (GCTs) are relatively common in the appendicular skeleton and frequently occur in the epiphyseal region. They rarely occur in the spine, and when they do develop in the axial skeleton, it is usually in the sacrum. Giant-cell tumours are osteolytic tumours comprising about 5% of primary bone tumours<sup>1</sup> and less than 5% of primary bone tumours in the spine.<sup>2</sup> They occur primarily during the second, third and fourth decades of life, with a slight predominance among women. These lesions tend to affect the vertebral body and are locally aggressive. Metastases may occur, usually in the lungs. Multifocal occurrence is largely uncommon. Patients commonly present with localized pain and tenderness. Radicular and neurologic symptoms due to compression of nerve roots and the spinal cord may occur when the tumours become large.<sup>3</sup> Bladder, bowel and sexual dysfunction may occur as a consequence of nerve root involvement in patients with tumours of the sacrum.

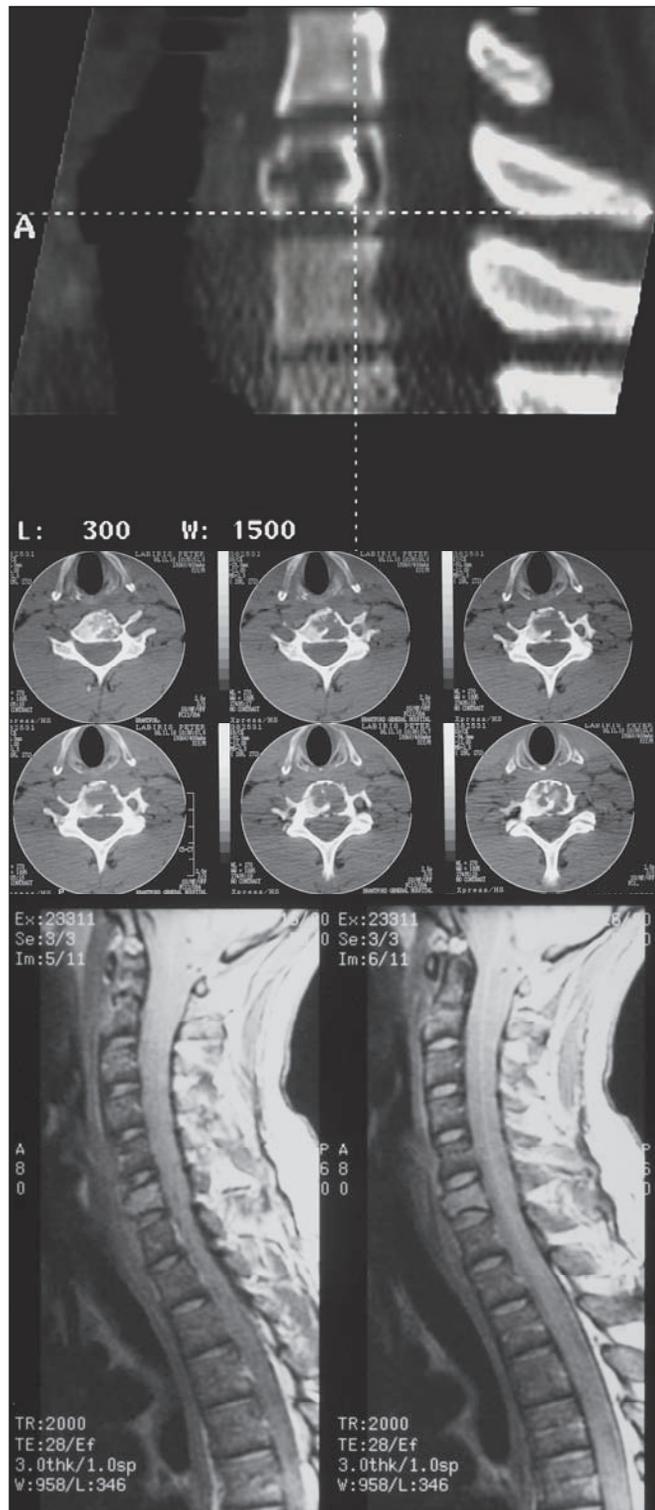
As with other spinal column tumours, management of GCTs presents a peculiar challenge. The difficulty lies in the proximity of critical structures and the propensity for the tumours to recur locally.<sup>4</sup> The mainstay of treatment is excision with wide margins. However, because these lesions can grow to a large size and compress neurovascular structures, wide excision is not feasible without the risk of substantial morbidity. Therefore, intralesional excision in conjunction with radiation therapy, with or without preoperative tumour embolization, is the recommended treatment option as there is less morbidity and reduced incidence of recurrence.<sup>5</sup>

**CASE REPORT**

We present the case of an otherwise healthy 32-year-old man who fell while playing ice hockey. He presented to the emergency department with neck pain. He had no neurologic deficit and no other injuries, and he denied any history of neck pain. Radiographs taken in the emergency department confirmed a fracture of the sixth cervical (C6) vertebra, with an obvious lytic destructive lesion. We admitted him to hospital and fitted him with a rigid cervical collar before sending him for further scans. An isotope bone scan showed increased uptake, confirming a solitary lesion at the C6 vertebra. The computed tomography (CT) scan showed a lytic destructive lesion confined to the C6 vertebra, associated with a vertebral body fracture. A magnetic resonance image (MRI) of his cervical spine revealed no significant cord compression (Fig. 1).

The patient's tumour was confined to the C6 vertebra. There was no soft-tissue extension. We considered giant-cell tumour, aneurysmal bone cyst, brown tumour, chondroblastoma and fibrous dysplasia as differential diagnoses. We concluded that our patient had a pathologic fracture at the

C6 vertebra. We proceeded with an anterior approach using a standard Smith Robinson technique from the right side.<sup>6</sup> Our patient underwent a C6 corpectomy and



**Fig. 1.** Preoperative (top) sagittal view of the computed tomography (CT) reconstruction of the C6 vertebra, (middle) axial CT scan of the C6 vertebra and (bottom) sagittal view of the magnetic resonance imaging scan of the C6 vertebra.

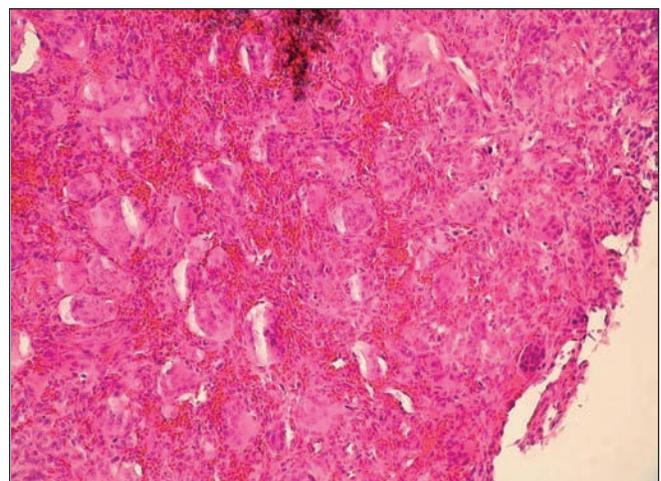
reconstruction of a defect with a tricortical iliac crest bone graft and an anterior titanium plate (Codman, DePuy Inc.). Using a magnification of  $\times 2.5$ , we achieved intralesional resection. Specimens sent for histology confirmed a giant-cell tumour with characteristic multinucleated giant cells (Fig. 2).

Postoperatively, our patient had no voice or swallowing complications. Blood loss was 1400 mL. He was mobilized with a neck brace, and we discharged him home by the fourth day after surgery. Postoperatively, solid fusion was achieved, the patient was free from pain, and he returned to work and sporting activities. He underwent annual surveillance consisting of clinical evaluation and imaging, including plain radiographs, CT scans and MRIs (Fig. 3).

After 7 years, there were still no recurrences, locally or systemically. The patient had adjuvant radiotherapy with fractionated 41.4 Gy of radiation starting 6 weeks after surgery to minimize the radiation effect in bone fusion and graft incorporation. He was fitted with a protective mask before radiation.

## DISCUSSION

Giant-cell tumours are benign bone tumours with high risk for local recurrence.<sup>7</sup> Overall, the recurrence rate of GCTs in the appendicular skeleton is 25%–45%.<sup>6</sup> They most commonly occur at either end of the long bones, and only 2.5% occur in the spine.<sup>2</sup> Most spinal GCTs occur in the sacrum. Those in the cervical spine are particularly rare.<sup>8,9</sup> The recurrence rate in a study evaluating the outcomes of patients treated at 3 different institutions was 28% in 36 patients.<sup>4</sup> Patients with GCTs in the cervical spine typically present with axial or radicular pain<sup>2</sup> and neurologic symptoms, and fracture may occur. Our patient presented with traumatic fracture without prior axial neck pain. We believe this signalled early presenta-



**Fig. 2.** Histologic appearance of benign giant-cell tumour showing hypercellularity with numerous giant multinucleated cells and a population of mononuclear cells uniformly dispersed.



**Fig. 3.** Plain radiographs showing the postoperative (**top**) anteroposterior and (**bottom**) lateral views of the C6 vertebra and plate.

tion that may have explained his long survival and lack of recurrence. We recognized the tumour immediately, which was a great benefit to our patient.

A previous report has documented the case of a patient who sustained a traumatic fracture following a motor vehicle collision in whom a GCT at the seventh thoracic (T7) vertebra was recognized 3 months after presentation.<sup>10</sup> Initial assessment is usually with plain radiographs, which typically reveal a destructive osteolytic lesion.<sup>11</sup> An isotope bone scan helps to confirm solitary disease; multifocal disease is uncommon. Computed tomography and MRI serve to assess the local extent of the disease in bone and soft tissue. In our patient's case, Enneking classification determined a stage-III benign tumour.<sup>12</sup> Giant-cell tumours are known to be hypervascular tumours.<sup>13</sup> Although we did not have access to embolization, we believe it may serve to reduce bleeding and duration of surgery. Embolization can also be used to determine involvement of the vertebral artery and to facilitate balloon occlusion tests if vertebral artery ligation is anticipated. Angiography also locates major radicular arteries, which should be avoided to prevent spinal cord infarction.<sup>14,15</sup> How embolization affects survival has not yet been proven. In some circumstances, embolization may be therapeutic. However, recurrence is high in these situations. Although en-bloc resection with wide margins is indicated for long bones and sacral GCTs, the cervical spine location makes it difficult and risky owing to critical nearby neurovascular structures.

En-bloc resection with intralesional resection and adjuvant radiotherapy and preoperative embolization, if feasible, appears to be an acceptable form of treatment for GCTs located in the cervical spine. We found one case report in the literature in which en-bloc resection of a GCT in the cervical spine was performed, with no recurrence after 2 years.<sup>16</sup> Our patient has had a 7-year survival without recurrence or metastasis. Although cryosurgery and phenol ablation may have applications in GCTs in the long bones, the role of these treatment methods in GCTs in the cervical spine is limited by critical nearby neurovascular structures.

We planned radiation for our patient in collaboration with the cancer clinic. A protective mask to shield surrounding structures was designed for him. He had 41.1 Gy of fractionated mega voltage radiation administered in 23 fractions over a 4-week period. Radiation carries a risk of malignant sarcoma with a previously reported incidence of 11%–14%.<sup>17</sup> Mega voltage appears much safer in this regard.<sup>17</sup> Such risk factors should be discussed when counselling patients about radiation treatments. There is a clear advantage to using radiation in combination with adequate resection. This method achieves the lowest recurrence rates compared with radiation with incomplete resection and radiation alone.<sup>18</sup> We began radiation therapy 6 weeks after the bone graft to minimize the effect of radiation on bone healing.

CONCLUSION

We present a rare case of a GCT in the cervical spine. Our patient's case was compelling owing to his 7-year survival with no recurrence or metastasis. In the absence of a large number of patients with this rare disorder, isolated reports such as ours are helpful in comprehending and documenting GCT treatments and their associated outcomes. We believe that in our patient's case, early diagnosis, meticulous local disease control and adjuvant radiotherapy accounted for the long survival. We recommend biopsy, when possible, followed by selective angiography and embolization, especially when soft tissue is involved. This should be followed by resection and adjuvant radiotherapy to achieve adequate local disease control. In the future, antiangiogenic therapy might play an important role as an adjunct in the treatment of axial GCTs since its use in the treatment of recurrent skeletal GCTs is beginning to gain more clinical recognition.<sup>19,20</sup>

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**Competing interests:** None declared.

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