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Total En Bloc Spondylectomy of C5 Vertebra for Chordoma

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Study Design. En bloc resection of a chordoma of the C5 vertebra with wide surgical margins.

Objective. To present the surgical technique of total spondylectomy for a chordoma of the C5 vertebral body.

Summary of Background Data. Malignant bone tumors require wide resection. Wide resection by total en bloc spondylectomy is difficult or not feasible for malignant vertebral tumors of the cervical spine due to the peculiar anatomic complexity of this region, including the vertebral arteries and the neural structures. There are no previous reports of en bloc resection of cervical spine tumors with wide surgical margins.

Methods. Using staged posterior and anterior approaches, a total en bloc spondylectomy and spine arthrodesis was performed. En bloc excision of a C5 chordoma was achieved using a threadwire T-saw (Tomita and Kawahara, Kanazawa, Japan) with surgical margins free of tumor. The patient received postoperative adjuvant proton beam radiation therapy.

Results. The patient remains disease-free 9 years after the operation.

Conclusion. Total en bloc spondylectomy with wide surgical margins is feasible for malignant bone tumors of the cervical spine.

Key words: en bloc spondylectomy, chordoma, cervical spine, vertebrectomy, bone tumor, wide margin, malignant neoplasm, vertebral artery, proton beam radiation. **Spine 2007;32:E294–E299**

Chordomas are the most common solid tumors of the spine, accounting for one third of the primary malignancies of the spine.[1] Eighty-five percent of all chordomas occur in the sacrum or the clivus.[2] Only 6% of all chordomas affected the cervical spine.[2,3]

They are considered low-grade, slow-growing, locally invasive, indolent malignancies, that tend to behave aggressively in the region of their origin and metastasize late in their course.[2–6] There is general agreement that en bloc surgical resection with wide margins is the treatment of choice for these tumors because intralesional or marginal resection consistently lead to local recurrence, and there is no effective adjuvant therapy.[5–7]

Tomita *et al*[8,9] described an innovative surgical technique, termed "total en bloc spondylectomy," using a newly designed threadwire T-saw for malignant vertebral tumors in the thoracolumbar spine.

Fujita *et al*[10] attempted en bloc resection of a chordoma of the C5 vertebral body with ligation of the left vertebral artery and the use of the T-saw to cut the pedicles. However, the resection was intralesional because the tumor had invaded the neural foramen.

In 2 recent case reports,[11,12] en bloc resection of a multilevel upper cervical spine chordoma required sacrifice of the vertebral artery and unilateral C2–C4 nerve roots to achieve clear margins.

The current report describes a patient with a chordoma of the C5 vertebral body. The tumor was resected successfully by en bloc total spondylectomy with wide surgical margins using the T-saw. No neurovascular structures were sacrificed. An illustrative description of the surgical technique used in the current case is given.

Case Report

A 51-year-old woman was referred to the author's institution for evaluation of a C5 lesion. Her presenting complaint was neck pain that started 10 years ago but became markedly symptomatic during the last year. The pain was located in the posterior midline, and it radiated to the shoulders bilaterally. There was no weakness, numbness, or bowel or bladder dysfunction. Her past medical history was negative for any malignancy.

With the presumptive diagnosis of cervical spondylosis, she had undergone physical therapy and chiropractic treatments over the previous 3 months. Due to progressive pain, she had radiographic evaluation, which showed a C5 vertebral body lesion. A needle biopsy was done, and the histologic findings were suspicious for chordoma. Additional studies, including computed tomography (CT) scan and bone scintigram, ruled out any other osseous lesions.

On physical examination, there was midline tenderness over the spinous process of C5 and C6. Her cervical spine range of motion was normal. Neurologic examination was normal, including strength of the upper and lower extremities, sensation and deep tendon reflexes, and absence of pathologic reflexes.

Plain radiographs showed a poorly demarcated lesion of the C5 vertebral body (Figure 1). A CT scan showed a sclerotic lesion of C5 with an evident biopsy tract and no apparent soft tissue mass or canal encroachment. Chest CT was negative for metastatic disease.

Magnetic resonance imaging (MRI) showed diffuse signal abnormality within the C5 vertebral body, with

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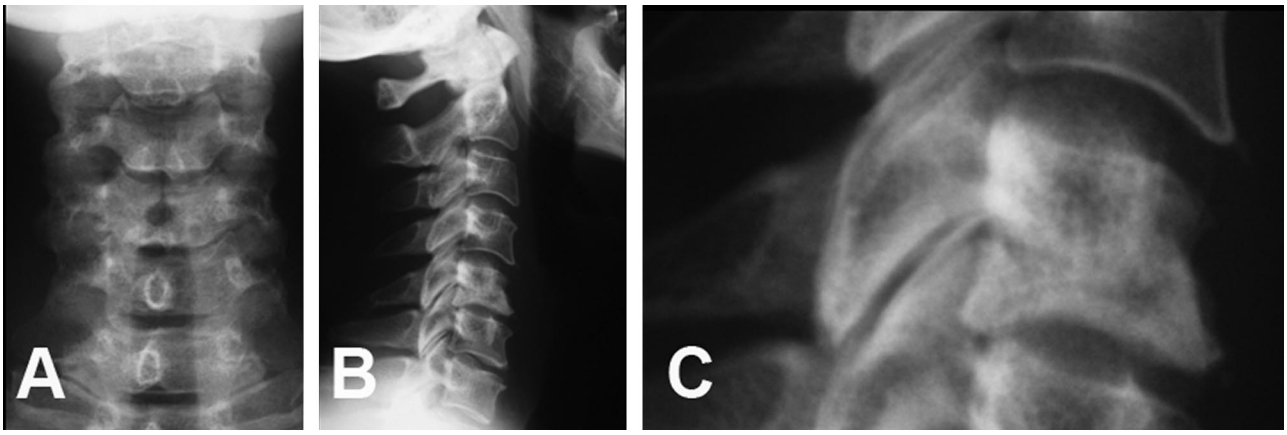

Figure 1. Anteroposterior (**A**) and lateral plain (**B**) radiograph of the cervical spine. There is a poorly demarcated lesion of C5 vertebral body shown on a magnified view of the lateral radiograph (**C**).

decreased signal on T1 and increased signal on T2 sequences (Figure 2). The lesion did not enhance with gadolinium contrast. There was no cord compression or foraminal involvement.

Bone scintigraphy and single photon emission CT showed well-localized increased uptake in the region of C5. Laboratory tests, including hematology, chemistry group, carcinoembryogenic antigen, and serum protein electrophoresis, were all within normal limits.

Surgical Procedure

A total en bloc spondylectomy was performed through staged posterior and anterior approaches (Figure 3). In the first stage, the posterior elements of C4–C6 were exposed subperiosteally, with dissection on both sides anterolaterally to expose the C4 and C5 nerve roots. The ligamentum flavum was excised between C4–C5 and C5–C6, and the C-5 lamina was removed in 1 piece by osteotomizing the bone at the junction between the lamina and lateral masses. The pedicles of the C5 vertebra were identified. Anticipating that once the lateral masses were excised the spine would fall into lordosis, drill holes were created in the lateral masses of C4 and C6. Using a medium-sized T-saw, the C5 pedicles were transected, and each lateral mass was removed en bloc. The posterior tubercles were excised to prevent entrapment of the vertebral arteries during removal of the vertebral body during the second stage. Posterior titanium plates and screws (Axis Fixation System; Sofamor Danek, Memphis, TN) were then secured to the spine, and bone grafts were secured to the spinous processes of C4 and C6.

In a second stage, 2 days later, the C-5 vertebral body was exposed anteriorly. The dissection was carried out to the tips of the anterior tubercles of the vertebra above and below, and the nerve roots were visualized and protected. Discectomies at C4–C5 and C5–C6 were performed back to the posterior longitudinal ligament. There was special attention not to enter the subchondral bone of the C5 body. The posterior longitudinal ligament was excised at C4–C5 and C5–C6. The vertebral arteries were dissected free of the C5 vertebral body and anterior tubercles. The body of the C5 vertebra was removed en bloc, and an iliac crest tricortical strut graft was placed in the vertebrectomy defect between the C4 and C6 vertebrae. An anterior plate was secured to C4 and C6 with 4 screws.

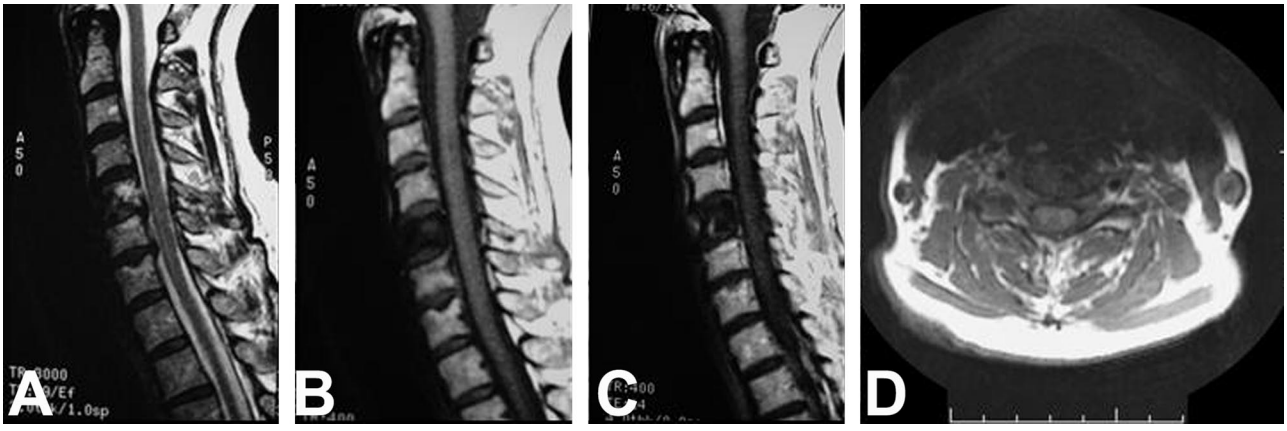

Figure 2. T2-weighted MRI showed diffuse increased signal within the C5 vertebral body (**A**). There was decreased signal in T1-weighted images (**B**). The lesion did not enhance with gadolinium contrast (**C**). Axial T1-weighted image shows no cord compression or foraminal involvement (**D**).

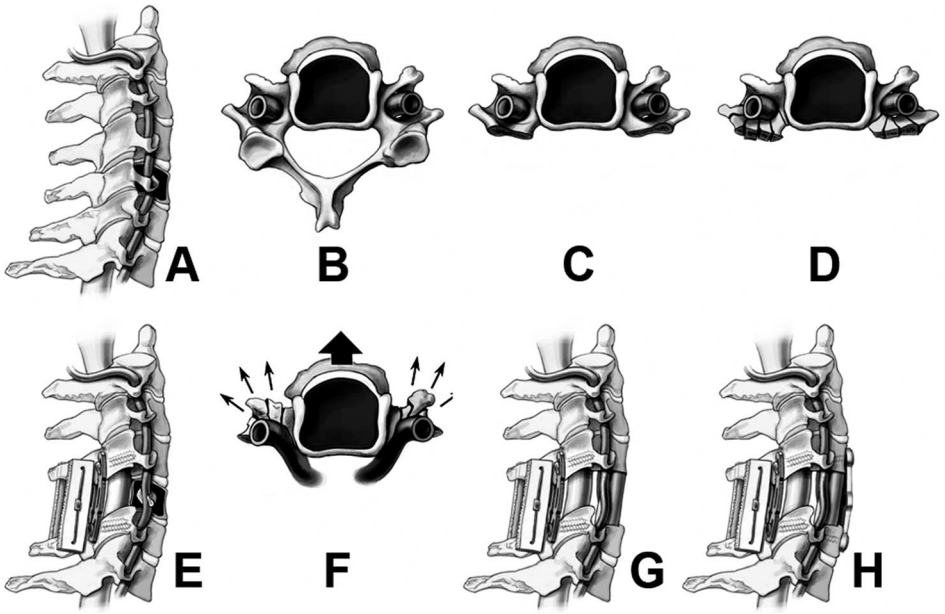

Figure 3. Drawings of the operative technique (**A**–**H**). The tumor (black) was contained in the vertebral body of C5 (**A**, **B**). A total C-5 laminectomy was performed removing the C-5 lamina in 1 piece (**C**). The lateral masses at C-5 were removed by cutting across the pedicle using a medium-sized T-saw. The remaining rim of bone posterior to the vertebral foramen was excised (**D**). Two strut grafts were placed posteriorly fixed with titanium cables to the spinous processes of C4 and C6 (**E**). Two 13-mm 3-hole titanium plates were fixed with 4–0 cancellous screws at the lateral masses of C4 and C6. Discectomies at C4–C5 and C5–C6 were performed (**F**). Dissection continued between the vertebral arteries and vertebral body and the joints of Luschka. Vertebral arteries were completely freed from the C5 vertebral body and anterior tubercles. The C-5 body was removed anteriorly. An iliac crest tricortical strut graft was placed anteriorly (**G**), and a plate was fixed with 4 screws to the anterior aspect of the vertebral bodies at C4 and C6 (**H**).

Histologic examination of the removed vertebra (Figure 4A) confirmed the diagnosis of chordoma involving the vertebral body. There were cells with voluminous clear cytoplasm that infiltrated the bony trabeculae. These "physaliphorous" cells had slight cytologic atypia (Figure 4B).

After surgery, there were no neurologic or wound-related problems. Three months after surgery, the patient received a continuous course of fractionated conformal precision radiation therapy. A total dose equal to 74.0 cobalt-Gy-equivalent was administered in 40 fractions. A partial dose of 41.4 Gy was given in 23 fractions of 1.8 Gy each using a 10-MV proton beam of a linear accelerator and employing a 3-field technique. An additional dose of 32.6 cobalt-Gy-equivalent was delivered in 17 fractions of 1.7 proton-Gy each using a range-modulated 160-MeV proton beam. The patient tolerated the treatment with no complications.

At her latest follow-up 9 years after her operation, the patient remains disease free. She had mild neck stiffness and no neurologic symptoms. Physical examination revealed an excellent range of motion of the cervical spine with mild restriction of rotation bilaterally. She was neurologically intact in the upper and lower extremities, and had no long tract signs. Plain radiographs showed a solid C4–C6 arthrodesis, and the instrumentation was in good position (Figure 5). There were mild degenerative changes at C6–C7. Flexion/extension views showed no instability. MRI revealed no evidence of recurrent tumor (Figure 6).

■ Discussion

It is generally accepted that en bloc surgical resection with a wide margin is the most appropriate management for chordomas.[5,10–14] Chordomas are not sensitive to chemotherapy, and only high-dose radiation therapy seems to slow the evolution of the disease.[15] Intralesional excision is associated with high recurrence rates.[5,7,11,15] However, in most cases of spinal chordomas, only marginal or intralesional resection is feasible because of the proximity of the tumor to vital anatomic structures, such as neural elements and the vertebral arteries.

Several studies describing the surgical excision of primary malignant or aggressive benign spine tumors have been reported, using combined anterior and posterior surgical approaches.[16–22]

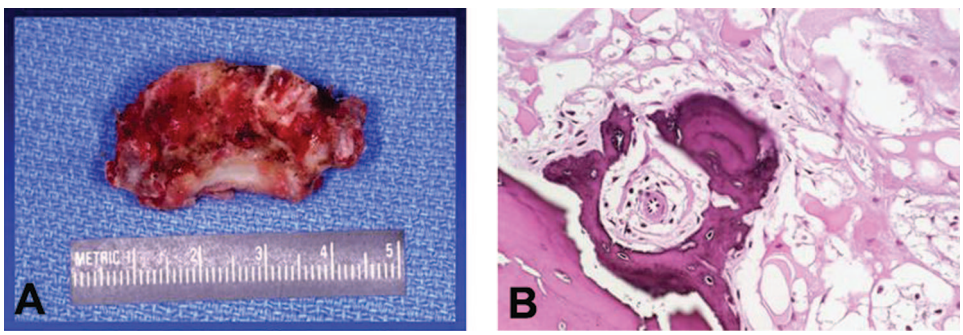

Figure 4. Macroscopic appearance of the removed specimen (**A**). High-power (80×) histologic appearance of chordoma showing "physaliphorous" cells with slight cytologic atypia (**B**).

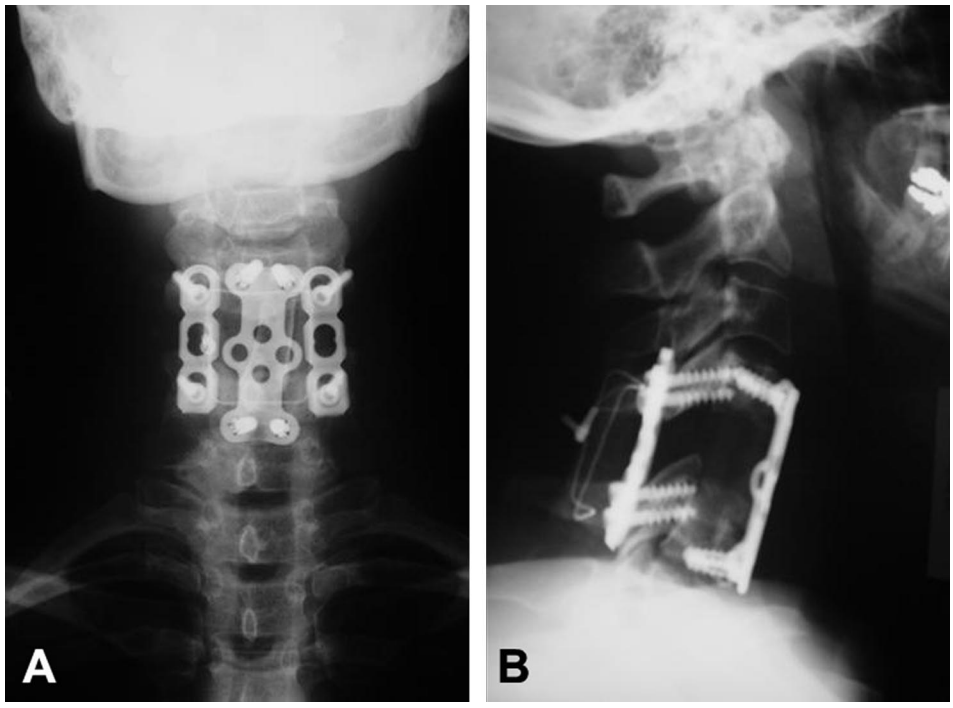

Figure 5. Anteroposterior (**A**) and lateral (**B**) views 9 years after surgery showing solid spine fusion and no instability. There were mild degenerative changes at C6–C7.

Total en bloc spondylectomy has been described to achieve complete tumor resection and oncologic cure of primary malignant vertebral tumors of the thoracolumbar spine.[9] The procedure is performed in 2 steps, consisting of en bloc laminectomy after bilateral pediculotomy using a stainless-steel threadwire saw (T-saw) and posterior spinal instrumentation (step 1), and en bloc corpectomy followed by anterior fusion with instrumentation (step 2).[8,9] The use of the T-saw is believed to minimize tumor cell contamination of the surrounding tissues or the surgical incision.[23,24]

Even with the use of the T-saw, wide en bloc excision of malignant tumors is complicated in the cervical spine due to the peculiar anatomic complexity of this region. The major risks of total en bloc cervical spondylectomy are injury to adjacent neural structures, the possibility of tumor contamination during pediculotomy, injury of vertebral arteries, excessive bleeding from the epidural venous plexus, infection, pseudarthrosis, and cerebrospinal fluid leakage.[9,10,22,25,26]

In the current patient, the tumor involved only the body of the C5 vertebra, and there was no soft tissue

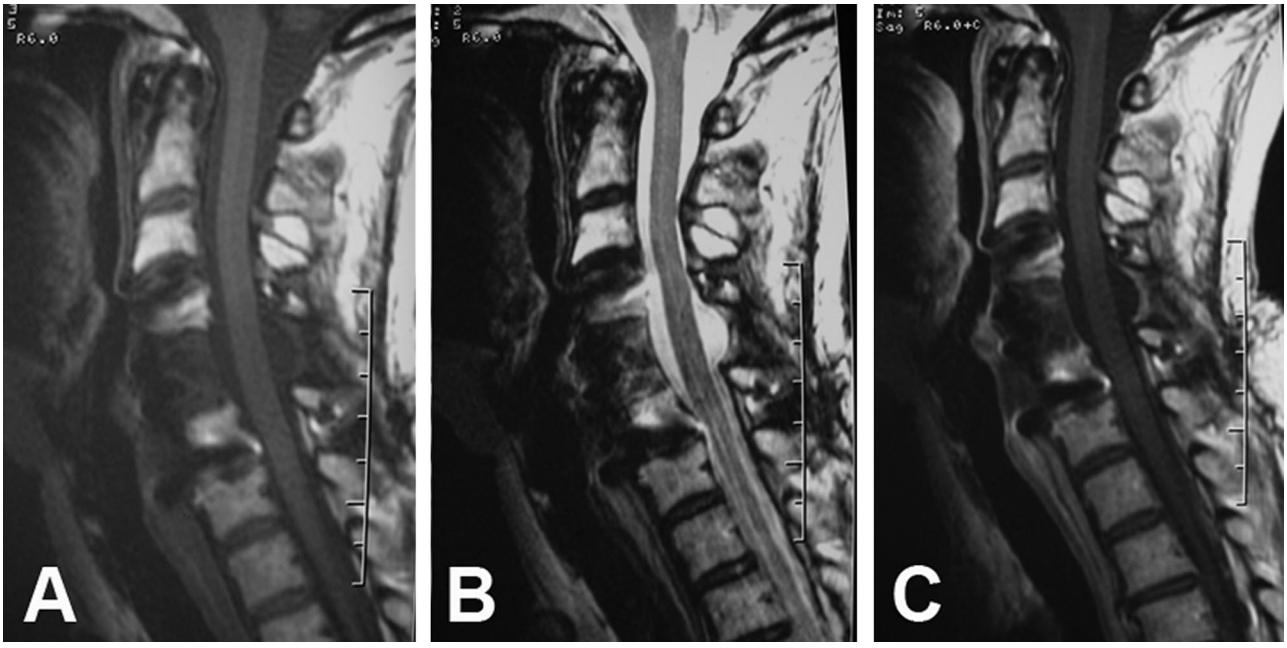

Figure 6. T1 (**A**), T2 (**B**), and with gadolinium contrast (**C**) MRI reveals no evidence of recurrent tumor.

extension or pedicle contamination. Total en bloc spondylectomy with wide margins was done with preservation of the vertebral arteries and nerve roots.

Cohen *et al*[22] described a technique for total en bloc spondylectomy for a C6 vertebra osteosarcoma that involved the vertebral body and the pedicles bilaterally with extension dorsal to the lamina and ventrally into soft tissues. The adjacent vertebral bodies were involved. The method involved separately staged posterior and anterior approaches for en bloc removal of the C5, C6, and C7 vertebrae with preservation of the vertebral arteries and nerve roots. However, the resection described in this case was intralesional. The patient had neoadjuvant and postoperative chemotherapy, and remained alive at least 10 months after the diagnosis.

For cervical spine tumors with soft tissue extension and involvement of the pedicles, total en bloc spondylectomy may require ligation of the vertebral artery.[10,25,27] Ligation of a vertebral artery should be performed only when preoperative angiographic study by a neuroradiologist demonstrates that the artery is nondominant, and temporary occlusion does not cause neurologic symptoms.[25] In a recent case report by Fujita *et al*,[10] a chordoma was located on 1 side of the C5 vertebral body. A special modification was made to allow the T-saw to be hooked inside the pedicle. The left vertebral artery was ligated, and troughs were made in the vertebral body and endplates of C6 and C4. However, the use of the T-saw resulted in an intralesional margin on the dura near the insertion of the left C6 root because the tumor had invaded the neural foramen at that level.

Rhines *et al*[11] reported en bloc resection of a multilevel cervical chordoma with C2 involvement that required a combined posterior-lateral/transmandibular approach with vertebral artery ligation, and C2, C3, and C4 nerve root sacrifice to achieve marginal resection at dural margin. In a recent report, Bailey *et al*[12] described en bloc resection of a seemingly unresectable C2 chordoma. Right hemi-spondylectomy of C2–C4 was achieved using posterior and anterior approaches in 3 stages. The tumor was removed with wide clear margins, except at the dura, where the margin was marginal. To achieve clear margins, the right vertebral artery and the C2, C3, and C4 nerve roots were sacrificed outside the margins of the tumor.

After total en bloc spondylectomy, the cervical spine is unstable and must be reconstructed. Rigid posterior reconstruction requires an autograft arthrodesis supplemented with plate or rod fixation secured with lateral mass or pedicle screws.[22] Autograft, allograft, spacers, or titanium cages can be supplemented with a plate for rigid anterior stabilization.[10,12,22,24] Chemotherapy has no current role in the management of chordoma.[14] Recently, imatinib mesylate has been found to have antitumor activity in patients with chordoma.[28] Based on imaging studies, evidence of disease stabilization and tumor necrosis was observed. Further studies are needed to determine the effect of imatinib therapy on outcomes.

The role of postoperative radiation therapy is poorly defined.[29] Conventional photon irradiation appears to result in poor local control in patients with macroscopic residual disease following surgery. We chose to administer radiation in this patient to increase the chance of cure; however, since we had achieved a wide margin, it may not have been necessary.[5]

The T-saw was used in the case to resect the lateral masses en bloc in an effort to reduce tumor spillage in case of subtle pedicle involvement. Post-resection histologic evaluation revealed that the tumor was isolated to the vertebral body of C5 and did not involve the pedicles or the lateral masses. In retrospect, it appears that the procedure could have been performed by removing the posterior elements piecemeal with similar results. Faced with a similar scenario, we would use the T-saw again, although it has not yet been proven that excising portions of bone involved with tumor in large sections is better oncologically than curettage or drilling with a burr.

■ Conclusions

En bloc resection of a chordoma of the cervical spine was performed with wide surgical margins and preservation of the vertebral arteries and neural structures. The patient is disease free 9 years after surgery.

■ Key Points

- Total en bloc spondylectomy with wide surgical margins is feasible in the cervical spine if the tumor is confined to the vertebral body.
- Total en bloc spondylectomy with wide surgical margins and adjuvant radiation therapy led to oncologic cure for a C5 chordoma.
- A cervical spine tumor confined to the vertebral body can be excised en bloc with wide margins without sacrificing neurovascular structures.

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