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En Bloc Resection of a Giant Cell Tumor in the Sacrum via a Posterior-Only Approach Without Nerve Root Sacrifice: Technical Case Report

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Abstract

BACKGROUND AND IMPORTANCE—Giant cell tumors (GCTs) are rare primary bone neoplasms. The best long-term prognosis is achieved via complete tumor excision, but this feat is challenging in the spine due to proximity of blood vessels and nervous tissue. When occurring in the sacrum, GCTs have been removed in an en bloc fashion via combined anterior/posterior approaches, oftentimes with nerve root sacrifice. The purpose of this article is to present a case of a single-staged, posterior-only approach for en bloc resection of a sacral GCT without nerve root sacrifice.

CLINICAL PRESENTATION—A 45-year-old female presented with intractable lower back and leg pain, saddle anesthesia, and lower extremity weakness. She underwent imaging studies, which revealed a lesion involving the S1 and S2 vertebral bodies. Computed tomography guided biopsy revealed the lesion to be a GCT. The patient underwent a posterior-only approach without nerve root sacrifice to achieve an en bloc resection, followed by lumbopelvic reconstruction.

CONCLUSION—Sacroctomy via a single-staged posterior approach with nerve root preservation is a challenging yet feasible procedure for the treatment of giant cell tumors in carefully selected patients.

Keywords

En bloc; Giant cell tumor; Nerve root; Posterior only; Sacrectomy; Sacrum

Giant cell tumors (GCT) are rare primary bone tumors and account for 7% to 10% of all primary spinal tumors.^{1,2} When affecting the spine, these tumors occur most commonly in

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the sacrum and typically in the third or fourth decade of life.³ GCTs are thought to originate from osteoclasts and are characterized by an expansile and locally aggressive nature. For this reason, en bloc resection is associated with the lowest local recurrence rates.^{4–9} Moreover, en bloc resections can be classified as wide if there is a clear margin of unviolated healthy tissue surrounding the tumor; marginal if the dissection is carried along the pseudocapsule enclosing the tumor; or contaminated if the pseudocapsule is violated or there is no discernible margin between pathologic and healthy tissue.¹⁰

Performing an en bloc resection in the sacrum often requires nerve root sacrifice.¹¹ Additionally, damage to the bowel and wound-related complications may also occur.¹⁰ The purpose of this article is to describe a high sacral amputation for en bloc resection of a GCT without nerve root sacrifice.

CLINICAL PRESENTATION

Presentation and Evaluation

A 45-year-old female presented with a 10-month history of progressive low back and right leg pain. During the past month she also developed saddle anesthesia, as well as bilateral leg weakness. On physical examination, she was uncomfortable in the sitting position, without being able to sit straight. Proximally, her strength was 5/5 in both legs, but strength was 4/5 distally. She had a positive straight leg test on the right. Sensory exam revealed decreased pinprick sensation in the S1–S2 distribution bilaterally, worse on the right.

A computed tomographic (CT) scan showed an expansile destructive lesion, appearing to originate from the S1 and S2 vertebral bodies (Figure 1). Magnetic resonance imaging showed a low-intensity lesion located at the S1 and S2 levels, with some peripheral edema (Figure 2). Additionally, there was expansion into the spinal canal, with significant compression of the cauda equina. The patient also underwent CT scans of the chest, abdomen, and pelvis to rule out metastatic disease; results were negative. The differential diagnosis included chordoma, GCT, multiple myeloma, and lymphoma. The patient underwent a CT-guided needle biopsy, which was consistent with a GCT. She additionally underwent angiogram and embolization. After a thorough discussion with the patient regarding the risks and benefits of treatment, the patient and surgeons opted for further surgical resection.

Operative Technique

The patient was placed in a prone position on a Jackson table. The patient's head was secured using a 3-point Mayfield head fixation. The operative field was shaved, prepped, and draped in the standard sterile fashion.

A posterior midline incision was made extending from the spinous process of L2 to the tip of the sacrum. Dissection was carried down in a standard fashion at L4 and L5, and an intraoperative radiograph was obtained to confirm the spinal level.

At this point, self-retaining retractors were placed in the operative field and attention was directed to the placement of instrumentation. Four 45-mm long, 6-mm diameter screws were

placed in the pedicles of L4 and L5 bilaterally. Using a high-speed burr, a complete laminectomy of S1, S2, S3, and S4 was performed bilaterally (Figure 3). The pedicle of S1 was then identified, and drilled bilaterally to allow exposure above the tumor mass. The pedicle of S3 was then identified and drilled below the tumor mass.

The thecal sac was then dissected free from the tumor mass, which was contained in the periosteal layers ventral to the sac. Vessel loops were placed around the S1–S5 nerve roots, which were reflected away from the tumor mass. After drilling the sacroiliac joints bilaterally, the ventral periosteum was identified after drilling a trough and incised (Figure 3). Ventral organ dissection was carried out using a #3 Penfield dissector (CareFusion, San Diego, California). The mesorectum was then visualized, and this was reflected away from the tumor mass in a caudal-to-rostral fashion.

In this fashion, the entire tumor mass was mobilized from the ventral attachment (Figure 4) and from the thecal sac. At this point, the tumor was delivered in 1 piece through the opening between the S1 and S2 nerve roots (Figures 5 and 6). In this case, while there was a distinct transition between tumor-involved bone and normal bone/bone marrow, there was not a distinct pseudocapsule, and the lesion was delivered en bloc with contaminated margins. Nevertheless, no discernible spillage of tumor occurred. The specimen consisted of a piece of dark red firm mass measuring $6.5 \times 5 \times 3$ cm. in size (Figure 7).

After removal of the tumor, the remaining bone edges were drilled using a high-speed diamond burr. Two 80-mm long and 8-mm diameter iliac screws were inserted bilaterally. A rod was then contoured to fit the lumbosacral lordosis. The rod was attached to the iliac screws and lumbar screws rostrally, and a lumbopelvic fixation was obtained from L4—pelvis. The operative field was then irrigated with copious amount of pulse lavage antibiotic irrigation. Afterwards, attention was directed to the fusion procedure.

Arthrodesis was achieved via drilling of the L4–L5 transverse processes, facet joints, and the dorsal ala bilaterally. Demineralized bone matrix and allograft bone chips were then placed bilaterally. Drains were then placed in the operative field: 2 in the subfascial space and 2 in the suprafascial space. The incision was then closed in a standard fashion. Total blood loss was estimated at 450 mL.

Postoperative Course

The patient tolerated the procedure well. She was seen by both physical and occupational therapists. The patient was discharged to home 7 days after her surgery with 5/5 strength in all muscle groups. Her incision was clean, dry, and intact and she was walking without difficulty.

Three-and-a-half years after surgery, the patient is pain free and walking without difficulty. She still has some mild numbness in the perianal region, but this is improved from preoperatively. There has been no need for further procedures, and there is no evidence of pseudoarthrosis.

DISCUSSION

GCTs consist of giant multinucleated cells thought to originate from osteoclasts.¹² Symptoms caused by these tumors are nonspecific and may include low back pain and neurological deficits due to nerve root compression. On CT scan, these tumors appear as expansile osteolytic lesions, and it is common to observe destruction of adjacent bone cortex.¹³ On magnetic resonance images, these lesions appear as lesions with low-to-intermediate signal intensity on T1- and T2-weighted images.

The treatment of choice is complete tumor excision, but performing such extensive resections in the spine carries a high risk of complications. When GCTs occur in the extremities, these lesions are usually treated with curettage and phenol placement in the resection cavity.¹⁴ However, this is not an option in the spine, as the phenol would most likely be hazardous to the nerve roots. In addition, the recurrence rate in the extremities is high, and re-excision may be a reasonable option. In the spine, re-excision puts the nerve roots at risk; therefore, an en bloc resection is preferred for tumor control.

GCTs may also metastasize, particularly to the lungs.¹⁵ With postoperative radiotherapy, there is also potential for malignant transformation.¹⁶ However, it remains unclear whether the radiation induces the malignant change, or the GCT tumors have an intrinsic malignant potential.

Historically, total and high sacrectomies have been performed via combined anterior/posterior approaches.¹⁰ This technique, which involves a laparotomy, allows for direct visualization and operative control of the rectum and vasculature anterior to the sacrum. This technique, however, also carries the risk of bowel injury and the inherent morbidity of abdominal surgeries. Based on the work by Fourney et al,¹¹ sacrectomies are classified based on the highest nerve root sacrificed—total sacrectomy (bilateral S1 and below or bilateral or unilateral L5), high sacrectomy (bilateral S2 and below or unilateral S1 and S2 and below), middle sacrectomy (bilateral S3 and below), low sacrectomy (bilateral S4 and below), and distal sacrectomy (S4 and above preserved).

Several factors should be taken into account in the decision-making process of how to approach sacral lesions. In general, anterior/posterior approaches are used if there is a large presacral component or if the tumor is in a high location, such as L5 or S1. Additionally, combined approaches allow for harvesting of a myocutaneous rectus flap or omental flap for closure.¹⁷ Anterior-only approaches are usually reserved for tumors in close proximity to the anterior sacrum, such as locally recurrent rectal cell carcinoma.¹⁸ In their description of an anterior-only technique, Fawaz et al commented: “potential disadvantages include the recognition that there is less direct control over the distal spinal canal equina [sic] elements below S1. A posterior only approach could afford more direct exposure to the terminal end of the dural sac with controlled ligation of exiting nerve roots distal to S1. We also recognize that a different surgical strategy may be required for patients with extensive direct bony invasion by tumor or in conditions that primarily involve bone itself.”¹⁸ Thus, posterior-only approaches are feasible when there is no rectal invasion by the tumor requiring diversion and

resection; the tumor is not above the L5/S1 disc space; and there is no iliac vessel involvement.¹⁰

Nerve root sacrifice is another consideration when planning these operations. For malignant tumors, such as chordoma, nerve root sacrifice should be considered due to the infiltrative nature of the tumor. On the other hand, when resecting benign tumors, nerve root preservation may be feasible. In the present case, although GCTs exhibit a locally aggressive nature, malignant transformation is uncommon, and the adjacent nerve roots were preserved.

In the present article, a posterior-only approach was performed allowing for en bloc tumor resection without nerve root sacrifice. Clarke et al¹⁰ reported outcomes of 36 patients treated via posterior-only approaches for various spinal tumors including chordomas, osteoblastomas, and sarcomas. The authors performed a variety of sacrectomies, including 2 total, 8 high, 9 middle, 12 low, and 5 distal. There was a 36% complication rate (13 patients), including 1 case of bowel injury. Notably, these cases involved nerve root sacrifice (with the exception of the distal sacrectomies).

Although posterior-only approaches avoid the need for a laparotomy, the structures anterior to the sacrum make access to the tumor more challenging. These procedures carry a relatively high morbidity rate, as mentioned previously. Potential complications after a posterior-only approach include bowel injury, unplanned nerve root injury, and wound-related complications. The latter perhaps represents the greatest challenge, given the fact that the surgical lesions are large and an omental flap or myocutaneous rectal flap is not available. When attempting a posterior-only approach without nerve root sacrifice, it is fundamental to consider both the primary tumor histology, as well as the location of the tumor.

CONCLUSION

Posterior-only approaches for en bloc resection of sacral tumors without need for nerve root sacrifice are feasible procedures in carefully selected cases.

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FIGURE 1.

A preoperative CT scan shows a lytic and expansile lesion involving the mid sacrum at the level of S1 and S2. Sagittal view (**left**) and axial view (**right**).

FIGURE 2.

A preoperative MRI scan shows a low-intensity lesion located at the level of S1 and S2. Sagittal view (**left**) and axial view (**right**).

FIGURE 3.

After a wide sacral laminectomy around the tumor site (Step 1), the sacral nerve roots are exposed and mobilized. Bone dissection circumscribing the anteriorly situated tumor is done by connecting the interrupted cuts (*) between the sacral foramina (Step 2 and 3). The periosteum is divided along the length of the sacral foramina (Step 4) to release the entire tumor specimen. Dissection of the periosteum lining the foramina also helps the surgeon establish the cleavage plane between nerve roots and bone. n., nerve. Reprinted with permission - Johns Hopkins University School of Medicine, Department of Neurosurgery - Ian Suk.

FIGURE 4.

The en bloc tumor specimen is gently mobilized by twisting it between retracted sacral nerve roots. n., nerve. Reprinted with permission - Johns Hopkins University School of Medicine, Department of Neurosurgery - Ian Suk.

FIGURE 5.

The tumor is delivered through a sacral window, without need for nerve root sacrifice. n., nerve. Reprinted with permission - Johns Hopkins University School of Medicine, Department of Neurosurgery - Ian Suk.

FIGURE 6. En bloc resection of the giant cell tumor. **Top**, the tumor is delivered in an en bloc fashion, leaving the S1 and S2 nerve roots intact (**Bottom**).

FIGURE 7.
Tumor specimen.