Sacral Nerve-Sparing Piecemeal Spondylectomy for Giant Cell Tumor of Bone in the Sacrum: Surgical Strategy and Accurate Tumor Location Identification

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Giant cell tumor of bone (GCTB) in the sacrum is an uncommon primary tumor in a difficult-to-treat anatomical area without a widely accepted treatment strategy.

A 46-year-old woman with a 5-year history of buttock pain was referred to our hospital. Physical examination revealed tenderness on the sacrum with inconclusive neurological symptoms. Image analysis revealed a large sacral tumor located at S1 to S5 with bone lysis (Fig. 1a, b), extending to the ventral and dorsal side of the sacrum (Fig. 1d, e). Needle biopsy revealed GCTB. Denosumab (120 mg) was administered five times on days 1, 15, 29, and 57, and before surgery[1]. This shrank the tumor, including the extraosseous tumors, and ossified the osteolytic portion of the bone without any apparent adverse events (Fig. 1c, f). Furthermore, tumor vessels were embolized a day before the surgery. She underwent piecemeal total spondylectomy of S2 to S5 and partial spondylectomy of S1, preserving all sacral nerves except left S4, which was accidentally severed during the surgery. Paravertebral muscles were flipped from the caudal side to expose the lamina and ossified tumor surface[2] (Fig. 2a, b, e). The iliocostalis muscle was also osteotomized at the iliac attachment and flipped along with the bone fragments (Fig. 2c, e). Double iliac screws were inserted bilaterally in the osteotomized surface. We created a red-colored CT image of the tumor with an approximately 5-mm margin, which was superimposed on the intraoperative CT image to confirm the exact location of the tumor using a navigation system (Fig. 3c). After removing the posterior lamina, S1 to S4 nerve roots were secured (Fig. 3a), and residual S2 to S5 pedicles were cut longitudinally using ultrasonic bone scalpel (Fig. 2d). Moreover, the alar of sacrum, including partial sarcoiliac joint, were removed. The residual sacrum body was removed, and complete excision was confirmed using the navigation system (Fig. 3b, c). All screws were connected, with L5/S interbody fusion and bone grafting to the SI joint (Fig. 4a, b). The wound was washed with a 0.35% iodine solution, and 1 g of vancomycin powder was locally administrated[3]. Flapped muscles were sutured to the anatomical position, and the gluteus maximus muscles were extended medially and sutured to cover the implants (Fig. 3d). Total surgical time was 9.4 hours, and total bleeding amount was 1,440 ml. For the first seven days postsurgery, blood glucose was controlled with insulin to prevent blood glucose levels from rising above 200 mg/dl[4]. Postoperatively, the patient experienced decreased perianal sensation, no relaxation of the anal sphincter muscle, and difficulty urinating on her own. Consequently, she was placed on intermittent voiding starting on the seventh postoperative day following a urologist's instruction. However, after two weeks postoperatively, she could urinate independently. She was discharged three weeks postoperatively, walking without any aid. After two years postoperatively, she continued to be disease free, returning to her previous work as a physician without adjuvant therapy.

In spinal GCTB, total *en bloc* spondylectomy (TES)[5] is recommended due to the high recurrence rate after simple intraregional curettage. However, TES of the sacrum involving S1 or S2 is not indicated[6] because sacrificing the sacral nerves can lead to bowel and urinary disorders. Complete removal of the GCTB, even piece by piece, is associated

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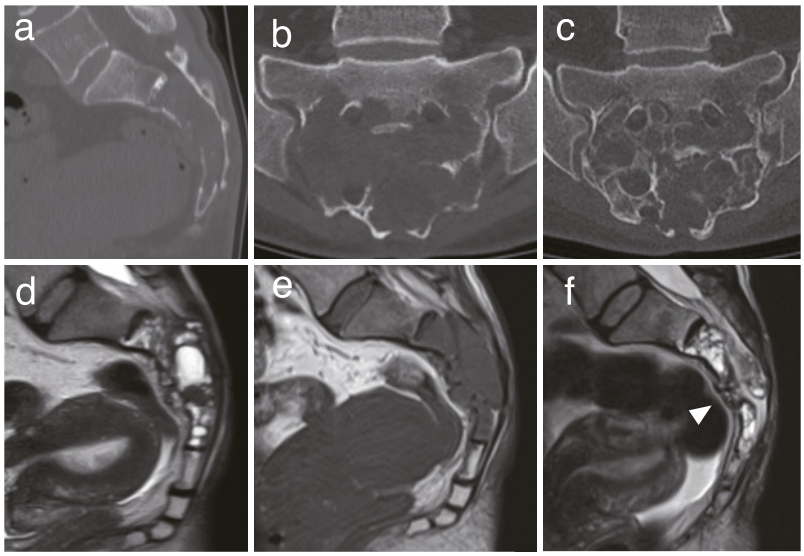

Figure 1. Image of the giant cell tumor of bone in the sacrum. Reconstructed sagittal (a) and coronal (b) computed tomography views revealing a large lytic lesion extending from S1 to S5. Magnetic resonance images (d and e) showing that the tumor extends to the sacrum's ventral and dorsal sides. After administering denosumab, ossification of the osteolytic lesion was clarified (c) and the tumor size decreased (f), including extraosseous tumors (white arrowhead).

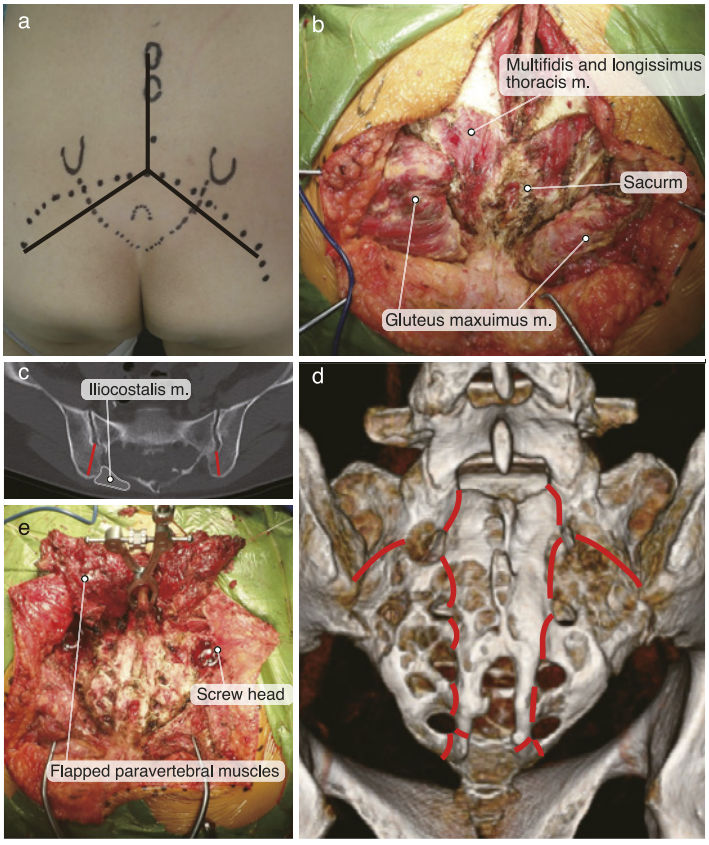

Figure 2. Skin marking and incision (solid black line) (a). The bilateral gluteus maximus, erector spine muscles, and sacrum are exposed (b). The posterior inferior iliac spine is osteotomized at the attachment of the iliocostalis muscles to adequately expose the sacroiliac joint and sacrum (c). The bilateral pediculectomy allows the removal of lamina and alar of the sacrum (solid red line indicates the cutting line) (d). Paravertebral muscles are flipped to expose the entire sacrum (e).

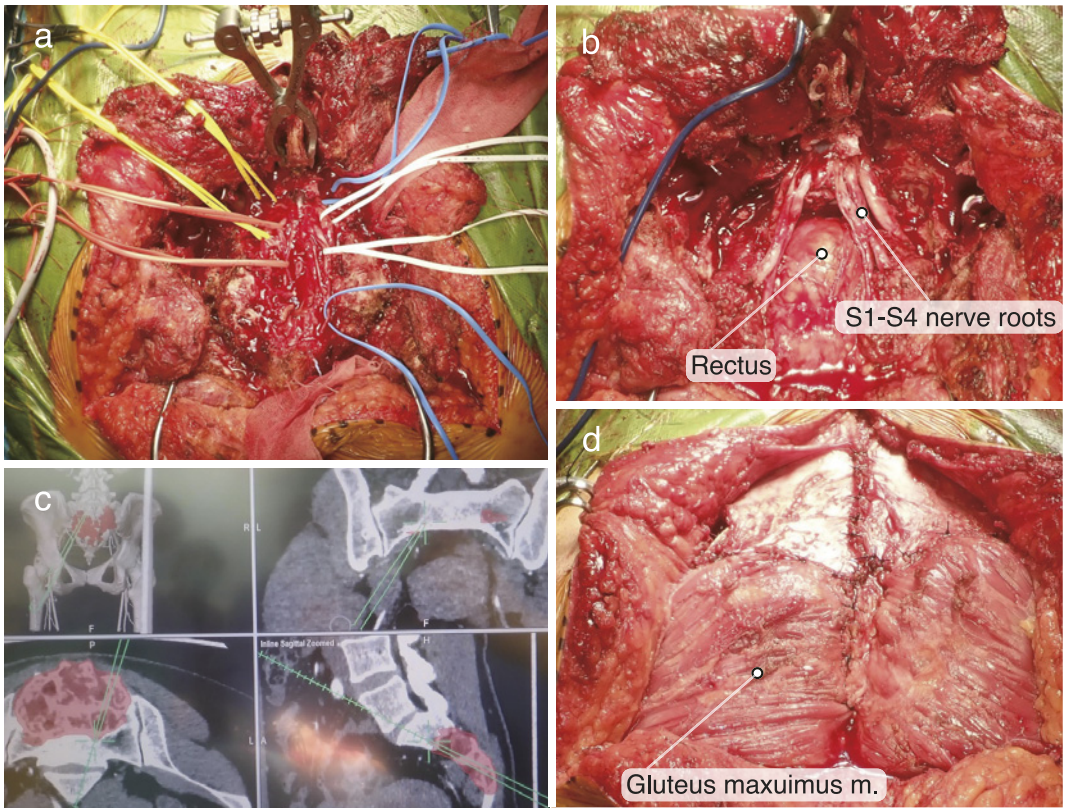

Figure 3. All nerve roots except for left S4 are secured (a) and complete piecemeal resection of the tumor is completed (b), as confirmed by computerized tomography-based navigation system (c). Bilateral gluteus maximus muscles are sutured medially to cover the rectus and spinal instrumentation (d).

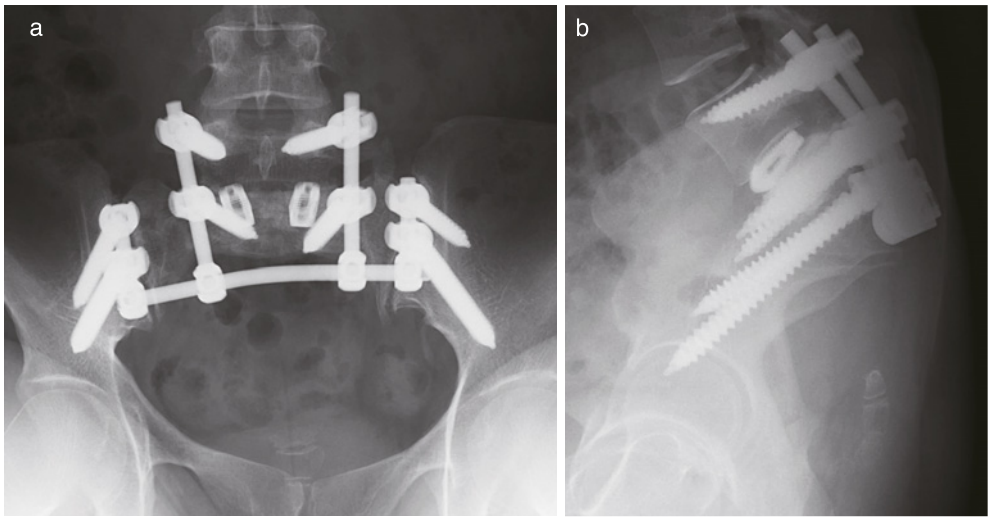

Figure 4. Anterior-posterior X-ray (a) and lateral X-ray (b) after surgery.

with lower recurrence[7,8]. GCTBs located in the sacrum have been observed to bleed an average volume of 3,000 ml, or more[6,9], and larger bleeding may result in incomplete resection, a risk factor associated with a high recurrence rate[9]. Although the effectiveness of adjuvant denosumab is highly debated[10], neoadjuvant treatment with denosumab has been reported to reduce the lesion and decrease intraoperative bleeding[10,11]. In this surgery, administration of denosumab and embolization may have decreased intraoperative bleeding. Iliac osteotomy is critical for sacroiliac joint exposure and aids in proper tumor resection. Soft tissue coverage by medial extension of the gluteus maximus muscle on both sides, together with iodine solution perfusion and local vancomycin powder application, can help prevent surgical site infection[3]. Furthermore, we prepared red-colored CT image with a margin in advance, which was useful for deciding the proper resection margin intraoperatively.

Although our current approach demonstrated positive re-

sults, the complexity of sacral GCT cases requires tailored, individualized treatment plans. Further research and collaboration among medical centers are necessary to accumulate substantial evidence supporting treatment standardization.

In conclusion, our case report demonstrates a successful surgical strategy for treating sacral GCT using preoperative denosumab, embolization, and nerve-sparing piecemeal spondylectomy. Furthermore, the patient's favorable outcomes with reduced bleeding, early recovery, and disease-free status after two years postoperatively are encouraging. However, cautiousness is essential, and well-designed prospective studies with larger patient cohorts and longer follow-up are needed to validate and refine the described combined treatment approaches. Advancing our understanding of this rare condition will improve treatment outcomes and enhance patients' quality of life.

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