Computer-Assisted Modified Mid-Sacrectomy for En Bloc Resection of Chordoma and Preservation of Bladder Function

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A 67-year-old woman presented for evaluation of severe coccygeal pain. The computed tomography scans and magnetic resonance imaging showed an asymmetric midline sacral tumor invading the right lower portion of S2. To preserve both S2 nerve roots and to obtain negative surgical margins, a modified mid-sacrectomy with an aid of a computed navigation system was performed. The sacral tumor was excised en bloc with negative tumor margins. Both S2 nerve roots were preserved and additional reconstruction was not necessary because of minimal resection of the sacroiliac joint. We report a case of a sacral chordoma which was excised en bloc with adequate surgical margins by a computer-assisted modified mid-sacrectomy. The computed navigation system may be a useful tool for tumor targeting and safe osteotomies in sacral tumor surgery via the posterior only approach.

Key Words: Sacral chordoma - Sacrectomy - En bloc resection - Computed navigation system.

INTRODUCTION

Surgical resection of sacral tumor is still a challenge due to anatomic complexity and difficulty in achieving wide surgical margins. En bloc resection with wide margins is recommended for sacral chordomas in particular because chemotherapy or conventional radiation therapy is not effective and the local recurrence rate is high. Since Nolte et al. first reported the successful clinical application of an image guidance system for lumbar pedicle screw placement in 1995, navigation technology has been widely applied in spine surgeries. However, most of the computer-guided spine surgeries have focused on accurate pedicle screw insertion and are still in experimental stage in the area of spinal tumor surgery.

We applied a computed navigation system for the intra-operative real-time localization of a sacral tumor and were able to resect the tumor en bloc with adequate surgical margins. In addition, we modified the known mid-sacrectomy technique (midsacral amputation) for en bloc resection of the tumor with partial invasion to the lower portion of S2, preserving both S2 nerve roots with minimal resection of the sacroiliac joint. Therefore, we report the case with a description of the operative technique.

CASE REPORT

History and examination

A 67-year-old woman was admitted with complaints of severe coccygeal pain. The coccygeal pain had slowly worsened over 1 year. She had undergone fusion surgery for foraminal stenosis at L4-L5 two months before admission. The neurologic examination showed a mild decrease in the anal sphincter tone and hypesthesia. There was no bladder incontinence. Magnetic resonance (MR) imaging revealed a midline sacral tumor involving S3 to the coccyx, with compression of the rectum. However, the mass invaded the lower margin of S2 on the right side (Fig. 1). Additional evaluation, including a chest and abdominal computed tomography (CT) and whole body bone scan revealed no distant metastases. Before surgery, a CT-guided needle biopsy was performed and the biopsy tissue was consistent...
midline skin incision was carried from L3 to the level of the coccyx. The dissection was carried laterally over the lumbosacral fascia to both iliac crests. The paraspinal muscles were elevated from L3 and the previous pedicle screws and rods were exposed. The paraspinal muscles were cut at the S2 level, leaving the distal portion covering the tumor.

After full exposure of the dorsal sacrum, a spine reference was attached to the right iliac crest by two pins. First, a paired-point registration was performed until the registration error was <5 mm. Surface registration was additionally performed until the registration error was <1 mm and the geometric constraint value reached 100%.

After the registration, the anococcygeal ligament and the levator ani muscles were divided from the coccyx. The avascular space between the rectosacral fascia (W aldeyer's fascia) and the presacral fascia was exposed to dissect the rectum from the tumor via the Kraske approach⁹. Laminectomies of S1-S2 were performed and S2 nerve roots were identified. The laminectomy was performed following S2 nerve roots to their exiting foramina. The thecal sac was doubly ligated below the S2 nerve roots with 2-0 silk suture ties and divided. The S3-S5 nerve roots involving the tumor were sacrificed.

First, the left lateral osteotomy was performed from the S2 foramen to the sciatic notch with a high-speed drill, protecting the S2 nerve root with a free elevator, with the usual method. However, during the right lateral osteotomy, the computed navigation system was applied to localize the tumor invasion to the lower S2 ala (Fig. 3). We outlined the tumor margin by a navigation probe and identified the osteotomy margin, which was a minimum of 10 mm from the tumor margin. The right lateral osteotomy was carried out by a high-speed drill with partial resection of the sacroiliac joint, including the posterior inferior iliac spine. During the osteotomy, the drilling location was frequently identified by the navigation system to prevent the wrong direction of the osteotomy and abrupt ventral perforation. Finally, the transverse osteotomy between the S2 foramina was carried out by the high-speed drill. The transverse osteotomy was also performed under navigation guidance. When the sacral osteotomies were completed, the distal sacrum became movable. The specimen was slightly tilted dorsally. Dissecting the ventral tumor from the rectum via the Kraske approach, we transected the lateral muscular and ligament attachments and the distal ends of the S3-S5 nerve roots. Finally, the tumor mass was resected en bloc with adequate surgical margins (Fig. 4). Both S2 nerve roots were preserved and additional reconstruction was not necessary because of minimal resection of the sacroiliac joint.

**Post-operative course**

Post-operatively, the patient was mon-
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itored in the intensive care and transferred to the general ward the next day. Acute bladder dysfunction and bowel incontinence were seen. Postoperative urodynamic study revealed a flaccid areflexic bladder. Seven days after surgery, she was able to ambulate. Bowel function was restored gradually. Her urologic symptom was improved, after the anticholinesterase was taken. The final histologic examination revealed a chordoma and all resection margins were free of tumor. The post-operative CT scan and MR imaging showed complete resection of the sacral tumor, including left lower S2 invasion and minimal resection of the right sacroiliac joint (Fig. 5). Twelve months after surgery, saddle anesthesia and minimal sphincter dysfunction was observed; however, urinary catheterization was not necessary. Motor function was intact.

DISCUSSION

Primary malignant sacral tumors are rare in adults. The most common primary tumors are chordomas and chondrosarcomas. These malignant tumors are relatively resistant to conventional radiation and chemotherapy therapy. Therefore, surgical resection is the primary treatment modality for primary malignant tumors. Because a negative surgical margin is the most important predictor of the tumor recurrence rate and survival, wide or marginal en bloc resection is the ideal treatment for primary malignant sacral tumors. However, according to the extent of tumor invasion, wide en bloc or extensive tumor resection may require combined anterior and posterior surgical treatment, resection of upper sacral roots, or lumbopelvic reconstruction with the risk of morbidity or mortality. Instead, marginal resection carries the possibility of tumor contamination or inadequate surgical margins, resulting in local recurrence. Thus, determining the adequate surgical margin is very important to minimize unnecessary resection and maximize preservation of neurologic function at the same time. Generally, planning the resection margin depends on the pre-operative CT scan or MR imaging. In symmetric midline tumor, the boundary of the tumor could be easily identified in operative field. However, there are some limitations in the intra-operative application of the pre-operative planning in acentric sacral tumors or asymmetric midline sacral tumors with irregular invasion. Specifically, in an en bloc sacrectomy with a posterior-only approach, which has been recently tried in sacral tumors, determining the adequate surgical margin is more difficult intra-operatively. Therefore, unnecessary wide resection is inevitable to achieve negative surgical margins with certainty. To avoid unnecessary resection and outline adequate surgical margins, we applied a computed navigation system in the mid-sacrectomy for en bloc resection of an asymmetric midline sacral chordoma.

The bladder’s parasympathetic efferent nerve supply originates from the sacral cord at S2-S4 and travels to the bladder via the pelvic nerve. The effect of parasympathetic stimulation is detrusor contraction. Also, somatic efferents originate from the sacral segment at S2-S4 and travel through the pudendal nerve.
and innervate the external urethral sphincter. In the current case, the midline sacral chordoma invaded the right lower ala of S2. For certain negative tumor margins in performing a high sacrectomy, the S2 nerve roots are sacrificed, causing profound bladder and bowel incontinence. Instead of a high sacrectomy, we modified a known mid-sacrectomy with a right perpendicular osteotomy to circle the tumor invasion. We outlined the tumor margin and determined the adequate resection margin with intra-operative real-time localization using a computed navigation system. To reduce mistake, a paired-point registration and surface registration was performed until the registration error was <5 mm and <1 mm. Then, the geometric constraint value reached 100%. Also, strong fixation of spine reference was done to reduce the mechanical errors. In addition, during osteotomies with frequent identification of drilling location by the navigation system, we prevented the incorrect ventral direction of the osteotomy and unexpected injury of the ventral organs. Finally, we resected the sacal chordoma en bloc with adequate surgical margins and preservation of both S2 nerve roots with a modified mid-sacrectomy. Additional reconstruction was not necessary because of minimal resection of the sacroiliac joint.

Recently, navigation technology has been widely used in neurosurgical fields. Also, the innovative technology has been popularly applied in spine surgeries. However, since Nolte et al. first reported the clinical application of an image guidance system for lumbar pedicle screw placement in spine surgeries in 1995, computer-guided spine surgeries have been focused on accurate pedicle screw insertion. Attempts have been made to apply navigation technology in other area of spine surgeries, e.g., ventral spine surgery, disc replacement, or tumor surgery. However, in spinal tumor surgery, application of the navigation system is still in experimental stages. With the exception of image-guided pedicle screw fixation, application of a navigation system for precise targeting of spinal tumors to avoid wide resection has been limited in resection of small benign bone tumors, such as osteoid osteomas. The application of a navigation system in sacral tumors has rarely been reported. Cho et al. reported a successful en bloc resection of an eccentric primary malignant tumor of the sacral ala with the aid of a computed navigation system. The current case is the first report of computer-assisted modified mid-sacrectomy for an asymmetric midline sacral chordoma. The computed navigation system was used in precise targeting and decision of the resection margin of a midline sacral tumor with asymmetric invasion. The tumor was successfully resected en bloc by a modified mid-sacrectomy. Although acute bladder dysfunction and bowel incontinence were seen temporarily which could be due to loss of parasympathetic outflow to the detrusor and/or afferent sensory information from the bladder and urethra. Because we preserved both S2 roots, the bladder dysfunction and bowel incontinence were improved immediately. The pelvic nerve and somatic pudendal nerve, which originate from sacral cord at S2-S4, are essential for preserving bladder and bowel function.

However, there are some limitations in the application of a computed navigation system for precise tumor localization in malignant spinal tumor surgery. First, most malignant tumors invade the surrounding soft tissue. The simultaneous navigation of both soft tissues and bone is still an unsolved problem in spine surgery, unlike navigated brain surgery. Second, in some spinal tumors, combined anterior and posterior surgery or anterior surgery is required, but the navigation of the tumor during the ventral approach is another problem to solve. Third, in malignant tumors involving multiple levels, such as metastasis, a navigational error occurs due to differences in vertebral alignment between the surgical and supine positions of the pre-operative CT acquisition.

Although image-guided surgery is still limited in spinal tumors, we suggest that computer-assisted surgery in sacral tumors is very useful, unlike tumors of the cervical, thoracic, and lumbar regions. The reasons are as follows. First, en bloc resection of sacral tumors via a posterior approach has only been recently tried, even in high or total sacrectomies. Second, the sacrum is a bony structure fused with 5 sacral segments and the coccyx. Thus, a navigational error based on surgical position is negligible. Therefore, computer-assisted tumor surgery is most feasible in en bloc resection of sacral tumors via a posterior only approach. We suggest that in malignant sacral tumors, the sacral nerve roots should be sacrificed and additional reconstruction due to unnecessary wide resection can be minimized by the application of a computer navigation system. In addition, we expect the application of a computed navigation system in spinal tumor surgeries will be more popular if merging a technique involving a CT scan and MR imaging can be developed in the spine, and intra-operative 3-dimensional CT scans can be acquired to minimize the positional error.

**CONCLUSION**

Image-guided surgery in spinal tumors is still in experimental stages. Specifically, computer-assisted sacral tumor resection has rarely been reported. The current case is the first report of computer-assisted modified mid-sacrectomy for an asymmetric midline sacral chordoma. The computed navigation system may be a very useful tool for precise tumor targeting and safe osteotomies in sacral tumor surgery via a posterior only approach.

**References**

1. Assaker R, Reyss N, Pertruzon B, Lejeune JP: Image-guided endoscopic spine surgery: Part II: clinical applications. *Spine (Phila Pa 1976)* 26: 1711-1718, 2001
2. Benevento BT, Sipuski ML: Neurogenic bladder, neurogenic bowel, and sexual dysfunction in people with spinal cord injury. *Phys Ther* 82: 601-612, 2002
3. Cho HS, Kang HG, Kim HS, Han I: Computer-assisted sacral tumor resection. A case report. *J Bone Joint Surg Am* 90: 1561-1566, 2008
4. Fourney DR, Rhines LD, Hentschel SJ, Skibber JM, Wolinsky JP, Weber KL, et al.: En bloc resection of primary sacral tumors: classification of
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5. Hsieh PC, Xu R, Scuibba DM, McGirt MJ, Nelson C, Witham TE, et al.: Long-term clinical outcomes following en bloc resections for sacral chordomas and chondrosarcomas: a series of twenty consecutive patients. *Spine (Phila Pa 1976)* 34: 2233-2239, 2009
6. Marshman LA, Friesem T, Rampersaud YR, Le Huec JC, Krishna M, Reddy GR: Significantly improved lumbar arthroplasty placement using image guidance: technical note. *Spine (Phila Pa 1976)* 32: 2027-2030, 2007
7. McLoughlin GS, Scuibba DM, Suk I, Witham T, Bydon A, Gokaslan ZL, et al.: En bloc total sacrectomy performed in a single stage through a posterior approach. *Neurosurgery* 63: ONS115-ONS120; discussion ONS120, 2008
8. Nolte IP, Visarius H, Arm E, Langlotz F, Schwarzenbach O, Zamorano L: Computer-aided fixation of spinal implants. *J Image Guid Surg* 1: 88-93, 1995
9. Onaitis M, Ludwig K, Perez-Tamayo A, Gottfried M, Russell L, Shadduck P, et al.: The Kraske procedure: a critical analysis of a surgical approach for mid-rectal lesions. *J Surg Oncol* 94: 194-202, 2006
10. Rajasekaran S, Kamath V, Shetty AP: Intraoperative Iso-C three-dimensional navigation in excision of spinal osteoid osteomas. *Spine (Phila Pa 1976)* 33: E25-E29, 2008
11. Ramamurthy R, Bose JC, Muthusamy V, Natarajan M, Kunjithapatham D: Staged sacrectomy—an adaptive approach. *J Neurosurg Spine* 11: 285-294, 2009
12. Rao G, Chang GJ, Suk I, Gokaslan Z, Rhines LD: Midsacral amputation for en bloc resection of chordoma. *Neurosurgery* 66: 41-44, 2010
13. Schwab JH, Healey JH, Rose P, Casas-Ganem J, Boland PJ: The surgical management of sacral chordomas. *Spine (Phila Pa 1976)* 34: 2700-2704, 2009
14. Tjardes T, Shafizadeh S, Rixen D, Paffrath T, Bouillon B, Steinhausen ES, et al.: Image-guided spine surgery: state of the art and future directions. *Eur Spine J* 19: 25-45, 2010
15. Van Royen BJ, Baayen JC, Pijpers R, Noske DP, Schakenraad D, Waisman PI: Osteoid osteoma of the spine: a novel technique using combined computer-assisted and gamma probe-guided high-speed intraleisional drill excision. *Spine (Phila Pa 1976)* 30: 369-373, 2005