INTRODUCTION
Sacral tumors are relatively uncommon. The majority of sacral tumors are metastatic tumors, although primary sacral tumors, such as primary bone tumors, neurogenic or congenital tumors, arising from the sacrum do occur.1-3 During diagnosis, sacral tumors are often missed at an early stage because of their indefinite clinical and unclear radiological characteristics. Due to their characteristics and growth patterns, these locally aggressive tumors often have huge masses and lead to complications.3 Computed tomographic (CT) scanning and magnetic resonance imaging (MRI) can aid in diagnosis in terms of the location, size, characteristics of these tumors, as well as in preoperative planning.

Most sacral tumors, except for metastatic tumors, are resistant to radiotherapy (RTx) and chemotherapy (CTx). Therefore, wide excision is typically the treatment of choice, and several surgical approaches and techniques have been described. Depending on the size and extent of the tumors, a single-stage posterior or a combined antero-posterior approach can be used.

The aim of this study was to review the management of sacral tumors, including symptoms, tumor features, perioperative management, surgical treatments, and complications.
MATERIALS AND METHODS

We retrospectively reviewed the records of 77 patients with sacral tumors who were treated between October 2011 and September 2019. Patients who underwent only tumor removal or surgical biopsy, as well as those with an intradural tumor, dermal or epidermal lesions, or recurrence after a previous surgery at other hospitals, were excluded. A total of 16 patients (6 males and 10 females) were analyzed. The mean age was 42.4 years (range, 16–79 years), and the mean follow-up period was 40.8 months (range, 12–79 months).

All of the patients underwent radiographs of the lumbosacral spine, including both sacroiliac (SI) joints, as well as CT scans and MRIs. The tumors were then classified by growth patterns according to guidelines for sacral tumors proposed by Wei, et al.3 (Table 1).

The clinical data of the patients, including age, sex, history, pathology, radiographs, treatment, recurrence, and prognosis, were analyzed. We obtained approval for this study from our Institutional Review Board (number: 3-2019-0128).

Surgical procedures

The surgical approaches differed between cases depending on the pathology, location, and extent of the tumor.

There is a notable difference between the upper and lower sacrum regarding spinopelvic continuity.1 Additionally, pre-sacral tumor protrusion of the tumor into the pelvic cavity and involvement of the SI joint and vessels surrounding and feeding the tumor are key factors in determining the surgical approach, specifically between a single posterior or a combined antero-posterior approach.

After deciding on the surgical approach, sacrectomy was considered. According to the system of Fourney, et al.,4 the sacrum is divided into three regions of the upper, middle, and lower sacrum by S1–S2 and S2–S3 junctions. Based on the tumor extension, en bloc resection of primary sacral tumors was classified into five types (Table 2).1,5,6

Combined antero-posterior approach

A combined antero-posterior approach is appropriate for type I tumors with presacral masses or type II tumors complicated by anterior masses greater than 5 cm. In type I, an en bloc resection with an anterior lumbosacral discectomy is useful. In lateral tumor invasion with involvement of the SI joint, en bloc resection and anterior SI joint removal are appropriate. Additionally, if the peritumoral surrounding vessels are complicated, an anterior approach is helpful for vessel ligation.

Surgery was performed in two stages. The tumor was approached anteriorly through an anterior midline transperitoneal or retroperitoneal approach.7 Recently, an anterior approach using laparoscope and retroperitoneoscope has been introduced.8 In most cases, it was difficult to accurately identify masses in the anterior; therefore, the tumor margin was explored and surgical vessel ligation was performed. Lateral dissection of the sacral ala allowed for identification of the lumbar trunk (L4–5) of the lumbosacral plexus. The SI joint was then identified lateral to these nerve roots, and bilateral partial ventral SI osteotomy was performed. The lumbosacral disc was exposed and removed along with the anterior aspect of the anulus fibrosis.

In stage II, via a posterior incision extending from L2 to beyond the coccyx, the posterior iliac crests, greater sciatic foramina, and sciatic nerves were exposed bilaterally, as well as the L3–5 spinous processes, facet joints, and transverse processes. An L5 laminectomy exposed the dural sac and cauda equine below this level. The sacral nerve roots were then divided, and the dural sac was amputated according to one of two methods: 1) amputation before closure, after which the dural sac was closed with a double layer of sutures, or 2) the dural sac was tied up several times first, after which the amputation was performed (Fig. 1). The remaining posterior L5–S1 intervertebral disc was then excised, and the posterior superior iliac spines were removed, facilitating cutting the bilateral osteotomes lateral to the ala of the sacrum and parallel to the SI joints, thus completing the osteotomy cuts made in these planes during stage I. Partial mobilization of the sacrum facilitated identification of the sacrospinous and sacrotuberous ligaments, which were then transected. The sacral nerve roots were divided as they exited the sacrum, protecting the sciatic nerves from injury. The entire sacrum along with the neoplasm was then removed en bloc.1,10–12

Single posterior approach

A single posterior approach is suitable for type II or III tumors with tumor extension to the anterior of less than 5 cm.

Table 1. Wei’s Classification (Sacral Neurogenic Tumor Types Classified by Growth Pattern)

| Classification          | Growth pattern                                           |
|-------------------------|----------------------------------------------------------|
| Type I                  | Confined to the sacral canal along with enlargement of the sacral canal |
| Type II                 | Forward out of sacral neural foramens, with formation of a huge presacral lump |
| Type III                | Spreads both anteriorly and posteriorly with formation of lumps anterior and posterior to the sacrum |
| Type IV                 | Confined to the presacral space, with no tumor present in the sacral canal |

Table 2. En bloc Resection of Primary Sacral Tumors

| Classification          | |
|-------------------------|------------------|
| Type I                  | Upper only, upper and middle, or upper to lower |
| Type II                 | Middle and lower |
| Type III                | Only lower       |
| Type IV                 | Eccentric lesions |
| Type V                  | Fifth lumbar vertebra is involved and has to be resected |
Reconstruction
In patients who underwent total sacrectomy, spinopelvic reconstruction was performed to facilitate early mobilization and better ambulation because of the spinopelvic discontinuity and instability.13,14 Fixation methods included various combinations of spinopelvic fixation (SPF), iliac screw fixation (ISF), and pelvic ring reconstruction (PR).

Two vertical L-shaped rods were positioned bilaterally in a manner allowing fixation to the L3–5 pedicles on each side according to the Galveston technique.15 Two to three cross-connecting rods were used to secure the vertical rods to each other. Distally, the vertical rods were directed laterally into the ilium between the two cortices. Both autologous and allogenic bone grafts were placed to promote fusion of the transverse processes and lamina from L4 distally to the medioposterior aspect of the transected ilium bilaterally. An allograft strut was used to close the space between the two ilia, and a bone fusion promoter and bone chips were added across the graft area to facilitate fusion of the entire defect. In cases where the dead space was wide, a gluteus muscle and skin flap were then utilized. In ordinary cases, a conventional muscle and skin closure was performed after placement of closed drains.16-18

Recently, 3D-printed implant reconstruction (3DIR) has been attempted. Therein, customized implants, which are fitted to a patient’s anatomy, minimize dead space and eliminate the need for additional reconstruction.19

RESULTS
Patient characteristics
The chief complaint of the patients was non-specific local pain. In terms of pre-operative motor deficits, 3 patients (19%) had leg weakness. Nine patients (57%) complained of bladder and bowel symptoms, such as voiding difficulty, urinary frequency or urgency, constipation, and residual sense after urination or defecation (Table 3).

Table 3. Clinical Characteristics and Preoperative Management of Patients with Sacral Tumor

| Case | Age/sex | CC | D (mo) | Motor | B/B | Size (cm) | G | Extent | Past history | CT guided biopsy | A/E | SB |
|------|---------|----|--------|-------|-----|-----------|---|--------|--------------|-----------------|-----|-----|
| 1    | F/70    | LP | 12     | Intact| UF, CoP, DAT | 9.8×3.5×7.5 | II | Middle | Thyroid Ca. | Chordoma | -/- | - |
| 2    | F/23    | LBP | 6      | Intact| UF, CoP | 8.5×8.0×9.8 | II | Upper | Pul. Tbc, NF | MPNST | +/- | - |
| 3    | F/39    | LP | 6      | Intact| Intact | 14.0×9.5×16.1 | III | Upper | Appendicitis | Osteosarcoma | +/- | - |
| 4    | F/38    | ButP | 1 | Both leg IV | Intact | 10.7×10.3×11.0 | III | Upper | - | Chordoma | +/- | - |
| 5    | F/41    | ButP | 18 | Intact | VD | 3.5×2.5×2.5 | II | Lower | - | Chordoma | +/- | - |
| 6    | M/29    | CP | 24 | Intact | Intact | 4.5×3.7×2.7 | II | Lower | - | - | +/- | + |
| 7    | F/58    | ButP | 12 | Intact | VD, CoP | 8.5×6.5×9.0 | III | Upper | - | - | +/- | - |
| 8    | F/58    | LBP | 24 | Intact | Intact | 3.0×6.4×5.1 | III | Middle | - | Schwannoma | +/- | - |
| 9    | M/27    | CP | 4 | Intact | VD, CoP, DAT | 8.5×4.5×4.3 | II | Middle | - | GCT | +/- | + |
| 10   | M/60    | CP | 3 | Intact | VD, CoP, DAT | 5.7×6.4×9.0 | III | Middle | HTN | Chordoma | +/- | - |
| 11   | F/79    | LBP | 4 | Intact | VD, CoP, DAT | 5.8×4.5×6.3 | II | Lower | HTN | Chordoma | +/- | - |
| 12   | F/16    | LBP | 4 | Intact | Intact | 3.9×2.9×3.9 | I | Upper | - | Osteosarcoma | +/- | + |
| 13   | M/55    | LBP | 36 | Intact | Intact | 4.5×4.8×5.3 | III | Middle | - | Chordoma | +/- | - |
| 14   | F/32    | LP | 7 | Intact | | 6.0×3.2×5.2 | I | Upper | - | GCT | +/- | - |
| 15   | M/21    | LP | 4 | Right leg III | VD, CoP | 8.8×9.0×9.0 | II | Middle | - | GCT | +/- | + |
| 16   | M/32    | LP | 12 | Left leg III | VD, CoP | 4.6×8.5×5.7 | II | Lower | - | - | +/- | + |

A/E, angiography/embolization; B/B, bladder/bowel; ButP, buttock pain; Ca., cancer; CC, chief complaint; CoP, constipation; CP, coccyx pain; CT, computed tomography; D, duration; DAT, decreased anal tone; DM, diabetes mellitus; f, female; G, growth pattern; GCT, giant cell tumor; HTN, hypertension; LBP, low back pain; LP, leg pain; M, male; Mo, months; MPNST, malignant peripheral nerve sheath tumor; NF, neurofibroma; Pul. Tbc, pulmonary tuberculosis; SB, sperm bank; UF, urinary frequency; VD, voiding difficulty.

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Pre-operative management
Before surgery, 13 patients underwent CT-guided biopsy for surgical and adjuvant therapy planning. Among possible complications, excessive blood loss is the most fatal and dangerous. Preoperative angiography was performed to identify highly vascular tumors. Subsequently, the decision between angiographic embolization or an anteriorly approached surgical ligation, or both, was made. In this study, 8 patients underwent preoperative angiography. For six of those patients, angiographic embolization was then performed using polyvinyl alcohol (150–250 μm or 250–300 μm) or coils via multiple feeding arteries from the internal iliac artery (Fig. 2).

Among six male patients, three were adults in their 20s who deposited their sperm in a sperm bank due to the risk of sexual dysfunction. The other patient in his 30s tried to deposit his sperm in a sperm bank, but was unable to do so, because ejaculation was impossible due to pain and it was thought that there would be no problems with sexual function after surgery.

Surgical management
Twelve patients underwent a single-stage posterior approach, and 3 patients underwent a two-stage antero-posterior approach. One patient underwent a two-stage anterolateral approach due to a chondrosarcoma located in an eccentric hemi-sacrum. The average blood loss was 5381 mL, and the average operation time was 8.24 hours.

In total, 5 patients underwent surgical iliac vessel ligation, all of which were performed after angiography to confirm a highly vascular tumor. In three cases, pre-operative embolization was performed first, but was not successful.

All patients who underwent a total sacrectomy and one patient who underwent a hemi-sacrectomy required SPF because the tumor extended S1 superiorly and the SI joint laterally. Because SI joint removal was required when the tumor was removed, they also required ISF and PR. A total of 2 patients underwent 3DIR with SPF and ISF, according to the method of Kim, et al.19

Two patients, one each with a high or middle sacrectomy, also underwent SPF. The patients’ tumors were large and extended to S1 but did not involve the root. Therefore, the upper part was removed only, and high or middle sacrectomy was done. In addition, because the SI joint was also involved, ISF was done in addition to SPF. For the patient who received a high sacrectomy, PR was also performed to prevent complications stemming from a pelvic gap after tumor removal. The procedure was performed by orthopedic surgeons using a universal locking system plate (Zimmer Biomet, Warsaw, IN, USA) and fresh-frozen femoral head allograft struts (Fig. 3).

A vertical skin incision was performed on most patients who underwent a posterior-only approach. However, for 5 patients with a large tumor extending up to S1 or laterally to the SI joint, a total or high sacrectomy was performed using an inverted T- or Y-shaped skin incision. The inverted Y incision allowed for easier tumor removal due to a wider operation field, compared to the inverted T incision, although the ensuing large void and severe skin defect required a muscle flap using the gluteus maximus to be created by plastic surgeons (Table 4).

Postoperative management
Seven patients (44%) complained of sensory changes, such as tingling sensation, numbness, and hypoesthesia. Six patients (38%) complained of post-operative motor deficits. Fourteen patients (88%) complained of bladder and bowel symptoms.

Of the patients with chordoma, five received adjuvant RTx.

Fig. 2 (A) Angiography for highly vascular tumors. There were multiple feeding arteries from the internal iliac artery (white arrow). (B) Angiographic embolization was performed using polyvinyl alcohol via a feeding artery from the internal iliac artery (white dotted circle). After angiographic embolization, there was no vascular flow from feeding arteries.
The patients with chondrosarcoma and osteosarcoma and one with giant cell tumor (GCT) received adjuvant RTx and CTx. One patient with neurofibroma received adjuvant CTx due to lung metastasis, not the sacral tumor. An additional patient with GCT received neoadjuvant CTx to decrease their tumor size. Another patient with Ewing’s sarcoma received palliative CTx. Three patients (19%) experienced post-operative recurrence (37 and 45 months for chordoma and 13 months for chondrosarcoma), and all of those patients expired (Table 5).

**DISCUSSION**

Sacral tumors are account for approximately 1–7% of all spinal tumors.26 Chordoma is the most common among the primary malignant bone tumors, and GCT is one of the most frequently seen benign lesions arising from the sacrum. Although GCT is a benign tumor, it is very vulnerable to local recurrence.21,22 Neurogenic tumors can also occur in the sacrum. Sacral neurogenic tumors, which comprise schwannoma and neurofi-
bromas, arise from the sacral nerve, grow along the bony neural foramen, and extend inside the sacral canal.4,23 In our study, chordoma was the most prevalent, followed by nerve sheath tumors.

En bloc resection is associated with decreased local recurrence and increased survival rates compared to intrasional resection. However, injury to the sacral nerve roots may occur intra-operatively, leading to post-operative neurological dysfunction.1,3,5,6,10 To do sacrectomy, dural sac amputation must be performed. In order to prevent postoperative cerebrospinal fluid leakage, careful watertight ties with non-absorbable silk suture material are required.24 As mentioned above, angiography is needed to identify highly vascular tumors. Pre-operative embolization can reduce intra-operative blood loss and time, increase tumor resectability, and improve visualization of the operative field. Even partial embolization may reduce intra-operative bleeding.25,26 In addition, patients who may wish to procreate in the future should be advised that sexual dysfunction may occur after surgery, allowing them to store their sperm in advance of the procedure.

Identifying tumor characteristics in advance through pre-operative CT-guided biopsy can also determine the surgical methods used and the appropriate treatment after surgery. However, during sacrectomy in a chordoma patient and biopsy in a resected tumor removed for intraoperative frozen, high-pressure exudate was observed coming from inside the tumor (Fig. 4). If a pre-operative CT-guided biopsy had been performed, we could not have ruled out the possibility that tumor cells had come out along the wound tract and invaded the surrounding area. Therefore, we have to ensure extensive tumor removal including the biopsy tract and skin. In addition to this method, if small chordoma is suspected based on pre-operative MRI, a total tumor removal without pre-operative biopsy is recommended to avoid tumor seeding.

The surgical methods to be used are determined in accordance with the size, location, and extension of the tumor. De-

| Case | Pathology     | Pain & sensory | Motor     | B/B     | RTx (T) | CTx (R) | F/U (mo) | Recurrence (mo) |
|------|---------------|----------------|-----------|---------|---------|---------|---------|-----------------|
| 1    | Chordoma      | ButP, HE(L5)   | Intact    | Foley, CoP, DAT | 29      | -       | 60      | 37, Expire      |
| 2    | Neurofibroma  | LP, LT         | Intact    | SV (Res), CoP   | -       | IE*     | 79      | DF              |
| 3    | Chondrosarcoma| LBP, LN        | Lt. ankle I | Foley, CoP, DAT | 30      | Cisplatin | 41      | 13, Expire      |
| 4    | Chordoma      | HE(L5)         | Both leg III | CIC, CoP, DAT  | -       | -       | 53      | DF              |
| 5    | Chordoma      | ButP           | Intact    | CIC, Fl        | -       | -       | 33      | DF              |
| 6    | Chordoma      | Intact         | Intact    | Intact         | 37      | -       | 23      | DF              |
| 7    | Schwannoma    | Intact         | Intact    | UI, CoP        | -       | -       | 18      | DF              |
| 8    | Schwannoma    | Intact         | Intact    | UI, CoP        | -       | -       | 17      | DF              |
| 9    | GCT           | Intact         | Intact    | Intact         | -       | -       | 22      | DF              |
| 10   | Chordoma      | CP             | Intact    | CIC, CoP       | 30      | -       | 67      | DF              |
| 11   | Chordoma      | ButP           | Intact    | UI, CoP        | 10      | -       | 55      | 45, Expire      |
| 12   | Osteosarcoma  | LBP, LN        | Lt. leg III | VD, CoP         | 28      | MAP     | 57      | DF              |
| 13   | Chordoma      | LBP            | Intact    | VD, CoP         | 20      | -       | 52      | DF              |
| 14   | GCT           | LBP, ButP      | Rt. ankle III | VD, CoP, DAT    | -       | Denosumab | 45      | DF              |
| 15   | GCT           | LBP, LT       | Both ankle I | Foley, CoP, DAT | 30      | Denosumab | 18      | DF              |
| 16   | Ewing’s sarcoma| LBP, HE (L3)  | Lt. leg III | Foley, CoP     | -       | VIDE    | 12      | OT              |

B/B, bladder/bowel; ButP, buttock pain; CIC, clean intermittent catheterization; CoP, constipation; CP, coccyx pain; CTx, chemotherapy; DAT, decreased anal tone; DF, disease free; Fl, fecal incontinence; F/U, follow-up; GCT, giant cell tumor; HE, hypoesthesia; IE, ifosfamide-epirubicin; LBP, low back pain; LN, leg numbness; LP, leg pain; Lt., left; LT, leg tingling; MAP, methotrexate-doxorubicin-cisplatin; Mo, months; OT, on treatment; R, regimens; Res, residual; Rt., right; RTx, radiotherapy; SV, self-voiding; T, times; UI, urinary incontinence; VD, voiding-difficulty; VIDE, vincristine-ifosfamide-doxorubicin-etoposide.

*Chemotherapy for lung metastasis.

Fig. 4. (A) On magnetic resonance imaging, a 4 × 4 cm round mass (white dotted circle) was located at S5. (B) Low sacrectomy and en bloc tumor resection were performed. (C) After en bloc resection, specimens were retrieved for intraoperative frozen biopsy. After biopsy, high-pressure exudate (white dotted circle) was observed coming from inside the tumor.
cisions are based on the level of sacrifice of the sacral root, which can have a direct effect on motor deficits, bladder and bowel symptoms, and sexual dysfunction that can occur after surgery. Therefore, it is optimal to save the root if possible. In this study, a single-stage posterior approach was used for a GCT patient with a large, hypervascular tumor covering the lower part of the S1 body and around the S2 root. During the stripping process to save the root, an uncontrolled iliac vessel injury occurred, causing the patient to undergo vessel ligation, resulting in increased operation time and bleeding. If a combined antero-posterior approach been planned for the surgery, the patient would likely have had a better result: the patient has had three surgeries, fortunately leading to a very good outcome overall. Indeed, Lee, et al. reported, in a case of presacral giant schwannoma, that an anterior approach could achieve total resection of presacral tumor without sacrificing sacral bone and sacral nerve root.

For tumors extending laterally to the SI joint, SI joint removal must be included when the tumor is removed. Six patients (38%) underwent SPF due to spinopelvic discontinuity and instability. SPF was performed in the 3 patients who underwent a total sacrectomy, followed by ISF and PR. An additional 3 patients underwent SPF and ISF; two of whom also received 3DIR.

In cases with a large tumor, long incision, and notable muscle dissection, the resulting void may be large after tumor removal, which can lead to wound dehiscence or necrosis. In our study, an inverted T or Y-shaped skin incision was performed in 7 patients. A gluteus muscle and/or skin flap was also utilized in these patients to prevent wound problems.

Adjuvant CTx or RTx should be considered as post-operative treatments according to the tumor pathology. In this study, patient prognosis was primarily affected by pathology and age. Three patients expired after recurrence. For the chondrosarcoma patient, the sacral lesion was well removed, but the tumor had heavily invaded the abdominal cavity such that only a near-total removal was possible. The remnant tumor was then subjected to adjuvant CTx and RTx, but the patient did not survive. Five of the 7 chordoma patients were under 60 years old, and the two others were over 70 years old. Unfortunately, despite the total sacrectomy and adjuvant CTx and RTx, both of the older patients experienced recurrence and expired.

The recommended treatment for sacral tumors is to remove as much of the tumor as possible. Surgeons should be careful of excessive bleeding during tumor removal, and therefore, pre-operative angiography and embolization are recommended. In addition, the type of sacrectomy should be determined according to the location and extension of the tumor, and to reduce post-operative complications, all possible efforts should be made to save up to the S2 root. In a systematic review paper, Zoccali, et al. reported that patients who underwent a sacrectomy maintained functionally normal ambulation in 56.2% of cases when both S2 roots were spared, in 94.1% when both S3 roots were spared, and in 100% with more distal resections. Normal bladder and bowel function were not present when both S2 were cut. When one S2 root was spared, normal bladder function was present in 25% of cases; in 39.9% when both S2 roots were spared, in 72.7% when one S3 root was spared, and in 83.3% when both S3 roots were spared. Abnormal bowel function was present in 12.5% of cases when both S1 roots and one S2 root were spared, in 50.0% of cases when both S2 roots were spared, and in 70% of cases when one S3 root was spared. If both S3 roots were spared, bowel function was normal in 94%
of cases. When even one S4 root was spared, normal bladder and bowel function were present in 100% of cases. Unilateral sacral nerve root resection preserved normal bladder function in 75% of cases and normal bowel function in 82.6% of cases. Motor function depended on S1 root involvement. In our study, Patients 14 and 15 had tumor invasion around the sacral root, and the pathology was GCT. Thus, if en-bloc resection was not performed, they were judged to be at high risk of recurrence, and the tumors were removed accordingly.

A limitation of our study was the relatively small number of sacrectomy cases. This number alone may not be enough to establish an optimal surgical strategy. In addition, there were differences in the treatment and follow-up periods depending on disease entities. Also, with the presence of several different operators for each individual, the resulting surgical strategy was not unified either. However, we believe that our data will contribute to and help in establishing theories as more data are added by other researchers.

If a tumor is large, a combined antero-posterior approach is recommended over a single-step approach. Spinopectineal re-construction must be considered following a total or high sacrectomy or SI joint removal, and for cases with a large incision or void following tumor removal, a muscle and/or skin flap should be considered. We have summarized our treatment algorithm in Fig. 5.

AUTHOR CONTRIBUTIONS

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