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Partial sacrectomy with patient-specific osteotomy guides

Mazda Farshad, Farah Selman, Marco D. Burkhard, Daniel Müller, José Miguel Spirig*

University Spine Center Zürich, Balgrist University Hospital, University of Zurich, Forchstrasse 340, 8008 Zurich, Switzerland

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ABSTRACT

Background: Chordomas are rare, locally aggressive, malignant tumors. Surgical resection with sufficient margins defines the outcome. However, the necessity for wide margins often leads to sacrifice of important neurological structures. 3D-printed osteotomy guides are a promising solution for precise execution of surgical resection. We present probably the first sacrococcygeal chordoma resection with 3D-printed guides.

Methods: The case of a 49-year-old woman with a sacrococcygeal chordoma, resected with help of 3-D pre-operative planning and patient-specific 3D-printed osteotomy guides, is reported in detail.

Results: A sufficient tumor excision could be performed successfully while sparing nerve root S4. The planed margin has been exactly maintained, as confirmed by histology. The patient demonstrated an excellent clinical outcome with no evidence of local recurrence.

Conclusions: 3-D pre-operative planning and patient-specific osteotomy guides can be used for planning and performing en-bloc surgical resection of sacral chordomas.

Background

Chordomas are relatively rare, malignant tumors of the spine, which arise from ectopic rests of notochordal tissue [1]. They are slow-growing and have a locally aggressive destructive behavior. Sacral chordomas commonly occur in individuals over 40 years of age, with men being twice as likely to be affected relative to women [2]. They are often localized in the clivus or the sacrum [3]. The most common symptom of a chordoma is local pain [4]. Nerve root or spinal cord involvement may further lead to neurological impairments (i.e. anorectal and urogenital dysfunction in sacral tumor location) [5]. These tumors often grow unnoticed as they do not cause any symptoms for a long time. Therefore, they are often diagnosed at an advanced stage, where significant bony destruction and soft tissue invasion has already occurred [3]. On average, a 12-14 month interval is reported between the onset of symptoms and the diagnosis of chordoma [4, 6]. Some are also found incidentally.

Percutaneous needle biopsy is the gold standard in the diagnostic workup if suspicion is raised according to CT and/or MRI. Additionally, CT and/or MRI can be used to determine the extent of chordoma lesions and facilitate surgical planning. Treatment of chordomas typically comprises of wide-margin surgical resection with or without radiation [4, 7, 8]. Chordomas have a low risk (5%) of metastasis [9]. However, the mortality is high, with an average life expectancy upon diagnosis of 5-7 years (untreated < 1 year) [4, 10]. The 5- and 10-year survival rates are 50-68% and 28-40% [11], respectively. The most important predictor of metastasis and mortality is local recurrence, which is clearly related to the extent of the initial resection margin [12–14].

Wide en-bloc resection is necessary to afford an opportunity for cure [4, 15, 16]. However, weighing up a wide tumor resection with consecutive neural damages versus a less extensive, nerve sparing resection with eventually increased risk of recurrence, remains the main challenge. In general, most surgeons plan for a resection margin of at least 2 cm, but exact suggested margin width varies in the literature [17]. Recent improvements in imaging and surgical techniques allow surgeons to plan and perform more precise osteotomy cuts. This, in turn, might allow tumor resections with maximum nerve sparing and sufficient surgical margins.

Patient-specific 3D-printed guides, planned and designed according to preoperative CT/MRI, allow custom-made cutting and drilling that helps the surgeon to perform an accurate and controlled procedure. 3D-printed guides are routinely applied for complex corrective osteotomies of the lower extremities, correction of malunions of lower and upper extremities and to optimize prosthesis alignment [18–20]. Due to their high precision and reliability, 3D-printed guides are being increasingly used in spine surgery, where they have found value in pedicle screw insertion [21, 22]. A recent report has further presented promising results in margin-respecting tumor resection in several anatomical regions [23].

To our best knowledge, we report the first case, in which a sacral chordoma was removed using patient-specific 3D-printed osteotomy guides and depth-limited osteotomes.
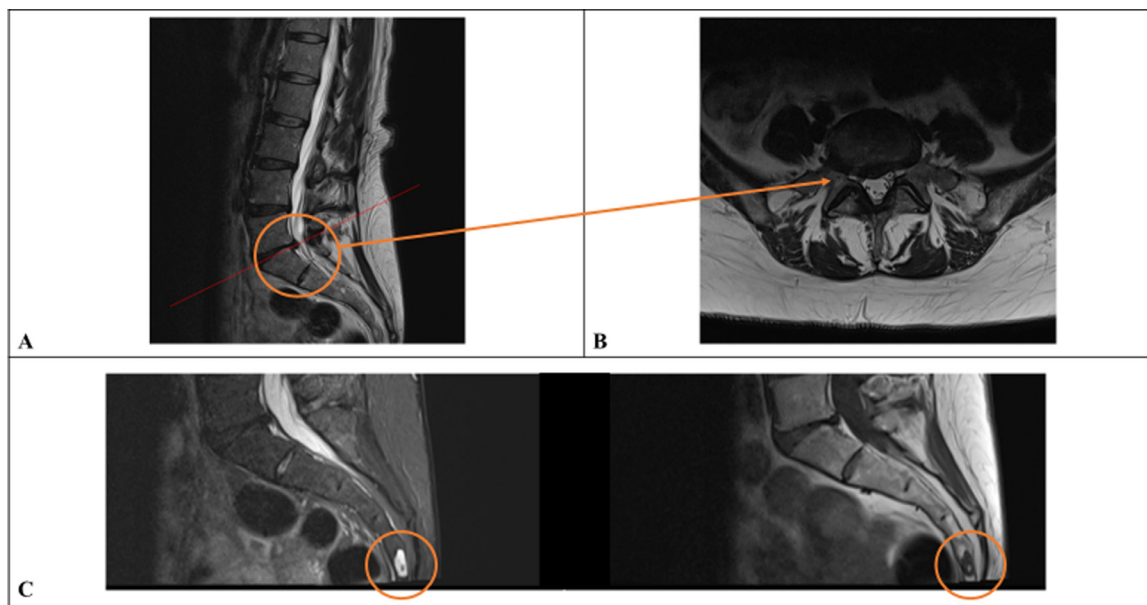

Fig. 1. Standard lumbar MRI performed for analysis of sciatic pain with incidental findings. A) Right lateral recess stenosis at L5/S1 explaining sciatic pain. T2 sequence, sagittal plane and B) transverse plane. C) Unexpected lesion in the 5th sacral vertebra. Left: hyperintense lesion in TIRM sequence, sagittal plane. Right: hypointense lesion in T1 sequence, sagittal plane.

Case presentation

A 49-year-old female initially presented with a history of sciatic pain on the right side for several years without any sensorimotor deficits and intermittent urinary and fecal incontinence. Her past surgical history was notable for cervical radiculopathy, which was successfully treated with two-level anterior cervical decompression and fusion surgery. Lumbar MRI was obtained to further analyze her sciatic pain, which showed some lateral recess stenosis at L5/S1 on the right side (Fig. 1A). However, in comparison to a lumbar MRI obtained 1 year prior, a new 15 mm-sized tumor in the 5th sacral vertebral body and another lesion in the coccyx were coincidentally detected. They showed morphologic characteristics suspicious for a chordoma (Fig. 1C). For more accurate imaging of the lesions, an MRI and CT of the sacrum were performed (Fig. 2). The following CT-controlled needle biopsy histologically confirmed the presence of a chordoma in the sacrum. The case was discussed at a multi-disciplinary tumor board at an academic medical center. Since there was no evidence of distant metastases in the thoracoabdominal PET-CT, systemic therapy was not indicated. For initial local therapy, there were options of surgical removal or proton irradiation. The patient decided for surgical resection. To spare the S4 root while respecting the minimum required margins, we decided to perform the partial sacrectomy using 3D-printed osteotomy guides. The patient agreed and a written consent was obtained to publish her case-related information and images.

Preoperative planning

Together with a team of biomedical engineers, a new form of 3D-printed guides were designed to respect both the tumor margins as well as neural structures. The guides were 3D-printed along with a 3D model of the sacrum using a SLS-printer (Selective Laser Sintering, EOS Formiga P396/P395) and sterilized before application in the operating room.

3D-surface models of the tumor as well as the sacrum were generated by segmentation and 3D-reconstruction of the MRI using MIMICS (Materialize, Leuven, Belgium). In addition to this MRI based model, the sacrum was again 3D-reconstructed based on the CT scan to allow a more detailed resolution of the bony surface of the sacrum. This was

important to reconstruct an exact bone-guide interface even though the tumor itself was not visible on the CT scan. The 3D surface models from both the MRI and CT were aligned to each other using the computer-aided design (CAD) surgical planning software CASPA (Balgrist CARD, Zurich, Switzerland). By doing so, a more resolute 3D surface model of the sacrum could be generated (based on the CT scan), which encompassed the correct size and location of the tumor model (based on the MRI).

A definite PSI guide development could then be realized including definition of resection margins, osteotomy planes as well as bone-guide interface areas.

The osteotomy guide comprised three blocks: one ground block (Fig. 3B), one on the ground block mountable osteotomy block for deroofing the sacral canal and bilateral exploration of nerve roots S4 (Fig. 3C), and one on the ground block mountable osteotomy block for the transverse osteotomy and the osteotomy of the vertebral body (Fig. 3D). Depth-limited osteotomes with 5 mm and 10 mm width were modified with a stopping element (Fig. 4). The planned transection gauges in the 3D-printed osteotomy guides stopped the osteotomes at the stopping elements after 80 mm, which made sure that no anterior soft tissue structures were to be endangered during the osteotomies.

Surgical technique

Patient was positioned prone on a Schaerer AXIS 500 table (Schaerer Medical AG, Münsingen, Switzerland) under general anesthesia. A midline skin incision over the sacrum to the coccyx was performed and the dorsal aspect of the sacrum exposed (Fig. 5A). Using electrocautery, the origins of the gluteus maximus and piriformis muscles were detached from the lateral aspects of distal sacrum. Caudal to the level of the planned sacral osteotomy, the anterior surface of the sacrum was bluntly dissected of the perirectal fascia and kept separated with laparotomy sponges (Fig. 5B).

The 3D-printed ground block was positioned at the level of S3 and fixed with Kirschner wires (Fig. 5C). The osteotomy for deroofing the sacral canal was then performed with the first osteotomy guide attached to the ground block (Fig. 5D). The depth-limited osteotomes were inserted into the osteotomy gauges of the guide until the osteotome was stopped from further penetration due to contact between the stopping
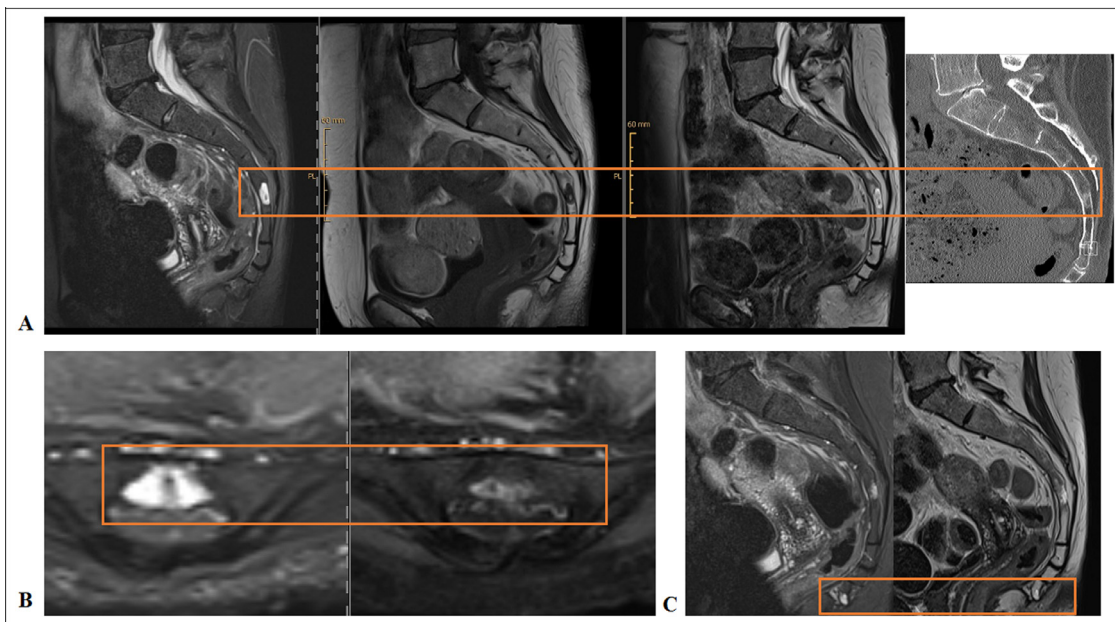
Fig. 2. Sacral MRI and CT with suspected Chordoma. A) Lesion in the 5th sacral vertebra, sagittal plane. First from left: hyperintense lesion in TIRM sequence. Second: hypointense lesion in T1 sequence. Third: hyperintense lesion in T2 sequence. Fourth: CT with slightly sclerotic lesion. B) Axial plane at 5th sacral vertebra. Left: hyperintense lesion in TIRM sequence. Right: slight contrast enhancement of the lesion in T1 sequence with Gadolinium. C) Lesion in the coccygeal tip, sagittal plane. Left: hyperintense lesion in TIRM sequence. Right: hyperintense lesion in T2 sequence. The lesions are well compatible with the presence of two chordomas.
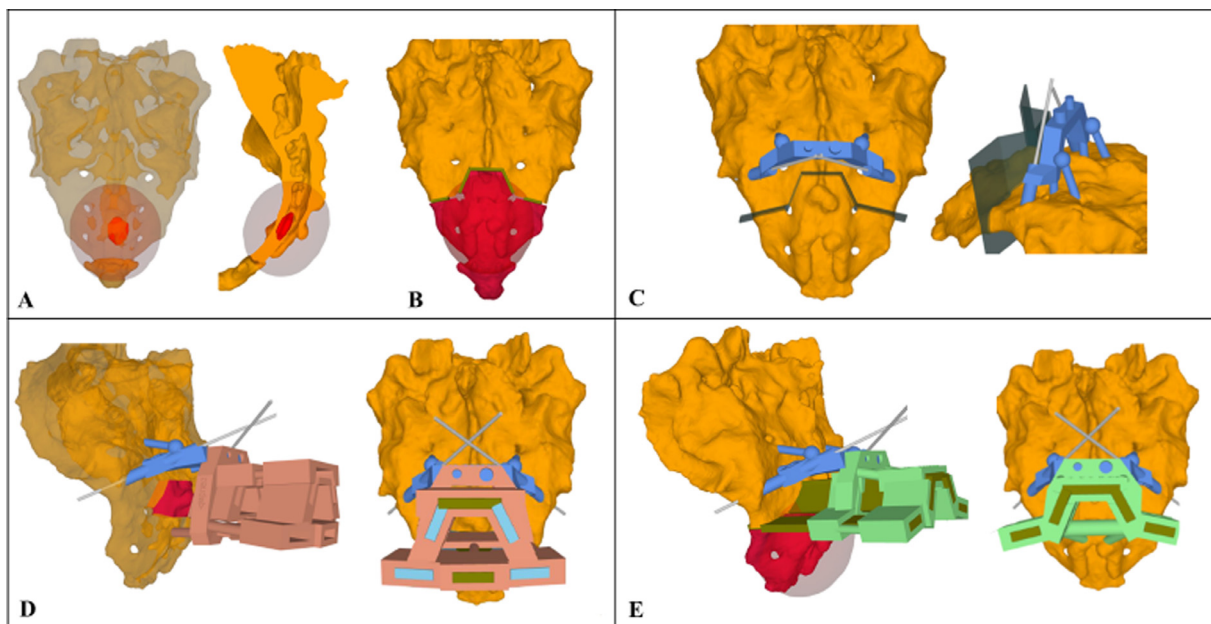

Fig. 3. Preoperative PSI planning. A) Sacrum (orange), Tumor (red) and 1.5cm resection margins (light red). B) Sacrum (orange), resection (red). C) Sacrum (orange), Ground block with 6 contact points and different attachments (blue), osteotomy plane (dark grey), 2 Kirschner wires (light grey) connecting the ground block with the sacrum. D) Sacrum (orange), Tumor (red), ground block (blue), Kirschner wires (light grey), Osteotomy guide for deroofing of sacral canal and bilateral exploration of nerve roots S4 (blush pink), connected with the ground block. Left: lateral view. Right: posterior view. E) Sacrum (orange), Tumor (red), ground block (blue), Kirschner wires (light grey), Osteotomy guide for transverse sacral amputation sub S4 (green).

element and the osteotomy gauge. After removal of the first osteotomy block, the osteotomized bone was removed and the sacral canal deroofed (Fig. 5E). No harm to the sacral nerves occurred. The nerve roots of S4 were bilaterally mobilized and spared. The remaining caudal nerves were ligated (Fig. 5F). Then, the second osteotomy guide was attached to the ground block and the transverse osteotomies of the sacral body were performed, while the S4 nerve roots were protected with a nerve root distractor (Fig. 5G). Again, deep penetration of the osteotome and

potential harm to the rectum and pararectal structures were avoided with the depth-limiting osteotomy technique. Then the caudal part of the sacrum was completely detached and carefully freed from soft tissue (Fig. 5H).

The partial sacrectomy and complete removal of the coccyx was performed with no harm to neural, vascular or visceral structures (Fig. 5I). Surgical time was 112 min and estimated blood loss was less than 100 ml.
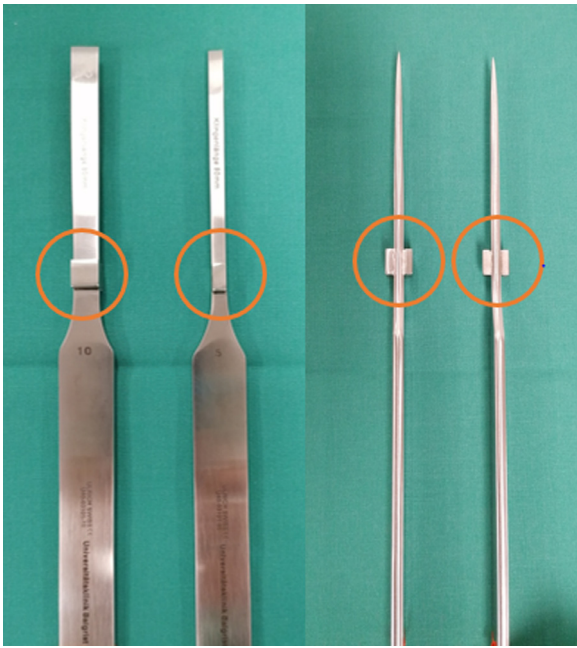
Fig. 4. Depth-limited osteotomes. 10 mm (left) and 5 mm (right) osteotomes were modified with a depth-limiting stopping element (orange circle) at a length of 80 mm.

Postoperative course

The immediate postoperative course was uneventful. The patient received usual oral osmotic laxatives such as polyethylene glycol and had

his first defecation on postoperative day five. There was no sign of fecal incontinence but some sacral sensory disturbance. The Foley catheter could be removed on postoperative day six with afterwards normal micturition of 350ml and a residual urine of 40ml under medication with a muscarinic antagonist (Fesoterodine). The patient was discharged home on postoperative day seven.

Postoperatively the patient was closely followed up by a urologist, a spine surgeon, and an oncologist. The histology of the resected tumor confirmed the entity of a chordoma and the compliance with the planed resection margins of 1.5 cm. In the 3 months postoperative outpatient visit, the patient could sit longer without pain and the initial postoperative sacral sensory disturbance was regressed. The last follow-up at 10 months postoperative showed no evidence of local recurrence on MRI (Fig. 6). In the first 2 postoperative years MR-radiological controls are planned every 3 months.

Discussion

Patient-specific guides are increasingly used in spine surgery [21, 22]. Tumor resection with guides showed promising results with high accuracy of resection at different anatomical regions [23]. We report here the first patient case in which patient-specific 3D-printed guides and depth-limited osteotomes were used for removal of a spinal chordoma by means of a partial sacrectomy with good results according to margin and nerve root preservation.

Chordoma resections and sacrectomies have been performed for over 70 years [24]. The challenge, however, remains to optimally plan and accomplish the required margin and at the same time to preserve neural, vascular and visceral structures and their function. Compared to conventional preoperative planning with CT and MRI, 3D analysis to plan 3D-printed osteotomy guides provide a better preoperative visualization and understanding of the tumor and its environment. This is crucial, as the anatomy of the sacropelvic region represents a major constraint
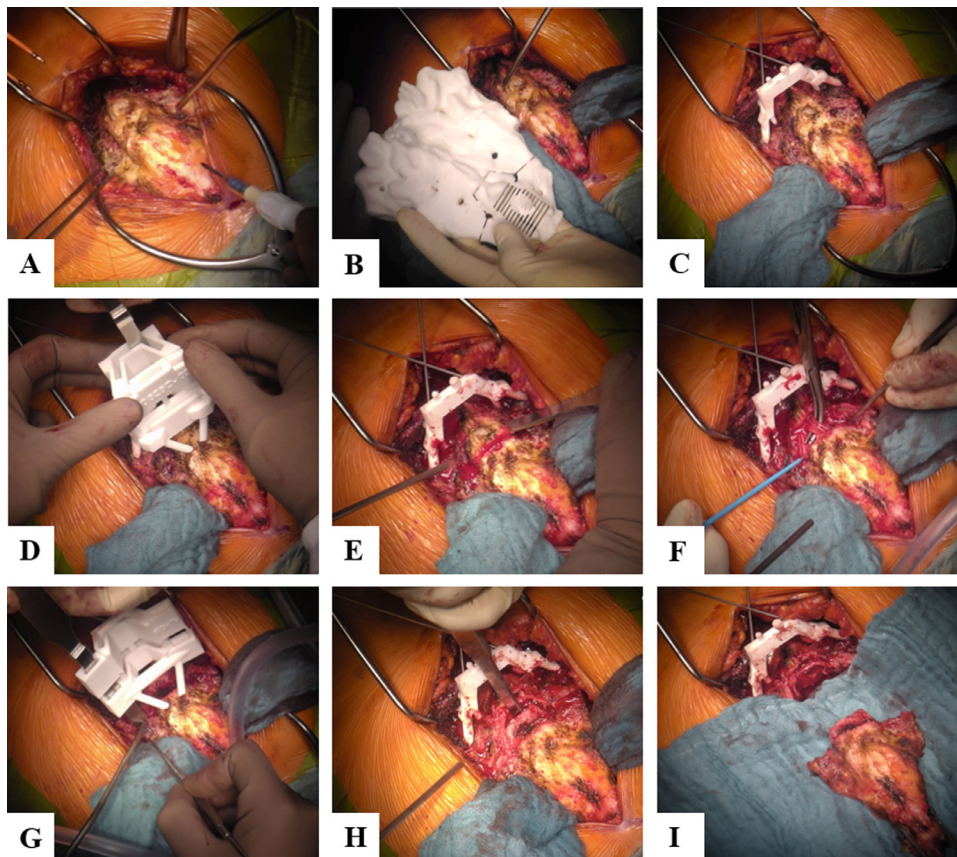

Fig. 5. Intraoperative photographs of PSI partial sacrectomy. A) Skin incision and dorsal exposure of the sacrum. B) Anterior sacro-visceral separation with a laparotomy sponge and referencing with patient-specific 3D-printed model. C) Ground block placement and fixation with K-wires. D) Osteotomy of dorsal aspect of sacrum through 1st osteotomy guide attached to ground block. E) Elevating the roof of the sacral canal after the 1st osteotomy. F) Ligation of sacral nerve roots with sparing of S4. G) Osteotomy of the anterior sacral wall through 2nd osteotomy guide attached to ground block. H) separation of the osteotomized distal sacrum by turning the chisel. I) Completed partial sacrectomy.
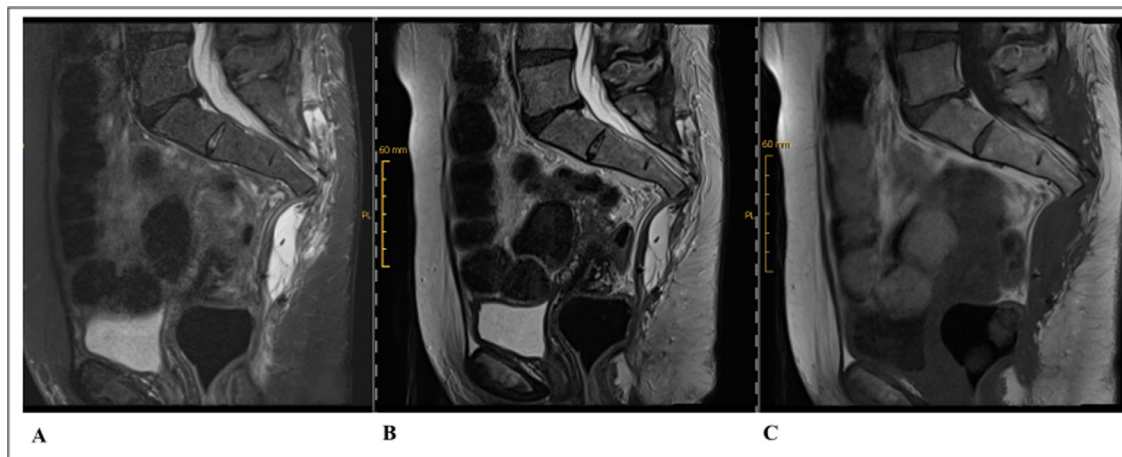
Fig. 6. Sacral MRI, 10 months postoperatively. No evidence of local recurrence in status post partial sacrectomy. A) TIRM sequence, sagittal plane. B) T2 sequence, sagittal plane. C) T1 sequence, sagittal plane.

to achieve local control in sacral chordoma treatment. Radical resection potentially requires sacrificing important structures, which result in permanent, life-changing functional sequelae and increased perioperative complications [25, 26]. One of the main concerns is to preserve the sacral nerve roots in order to prevent bowel and bladder dysfunction [27]. Achieving a sufficient en-bloc resection margin on the other hand plays a key role for the recurrence rate and therefore the long-term outcome [17, 28-30].

With the patient-specific 3D guide technique, we achieved an ideal compromise of both requirements. The S4 roots could be preserved and the surgical margin, which could be minimalized to 1.5 cm, was maintained as planned and confirmed by histology. This shows that the PSI technique delivers here a simple navigational tool to reliably adhere to the preoperative osteotomy plan, which is very important in maximally minimalized resection margins should it be necessary to preserve neurological structures. Strict compliance with the planned margin might be very difficult without any intraoperative navigation tool.

Other authors tried to achieve precise margins using robot-assisted multidisciplinary surgery with intraoperative neurophysiological monitoring [31, 32]. Long-term effects in terms of recurrence and survival haven't been investigated yet. Compared to robot-assisted surgeries, the use of intraoperative patient-specific guides is very user friendly as it does not need an expansive infrastructure and avoids an unfavorable learning curve [33]. The positioning of the 3D-printed ground blocks can be however challenging. It needs proper exposure of the bony surface. Soft tissue trapped under the guide could lead to major deviation of the osteotomy planes. The here presented depth-limited osteotomes can be used to ensure safety to soft tissue structures during the procedure.

Compared to robot-assisted surgery, our technique has advantages in terms of approach, blood loss, surgery and hospitalization time. Castiglione et al. reported a blood loss of 500 ml, surgery time of 300 minutes and a discharge after ten days for their combined anterior and posterior robot-assisted approach [31]. Chinder et al. recorded an average operation time of 360 minutes and median blood loss of 930 ml for a comparable procedure [34]. Our patient lost around 100 ml blood during 112 minutes of surgery with a posterior-only approach and was discharged after seven days. However, the cases are not directly comparable due to different size and location of the tumors.

Konakondla et al. presented a technique using intraoperative neuronavigation with preoperative MRI fused to intraoperative CT, creating a 3D tumor reconstruction in the operating room for intraoperative identification of bone and soft-tissue margins [35]. The fusion of preoperative MRI to intraoperative CT provides excellent visualization of the tumor, but it is rarely used because of unavailability and cost. Compared to usual radiological preoperative planning, intraoperative CT goes along

with higher radiation doses. In addition, the operation time can be extended through settings of the device (mean length of surgery 325 vs. 112 minutes).

3D-printed guides are preoperatively planed and produced, which is time consuming and may lead to a delayed surgery date. However, since sacral chordomas do not require immediate resection due to their slow growth, this disadvantage does not have a major effect on the therapy. After performing the right surgical approach intraoperatively, the guides are immediately ready for use, without extension of surgery or anesthesia time.

In our case, the technique used was safe and the intra-, peri- and postoperative course was excellent. We believe that patient-specific osteotomy guides are a valuable tool in sacral tumor surgery as demonstrated in our case. The question arises, whether this technique, as used for this case, can be adapted for tumors in other regions of the spine without endangering neural structures. Further investigations of this technique in other spinal regions are therefore warranted.

Conclusion

3D pre-operative planning and patient-specific 3D-printed osteotomy guides can be used for planning and performing en-bloc time-efficient surgical resection of sacral tumors with excellent accuracy and minimal blood loss.

Informed Patient Consent

The authors declare that informed patient consent was taken from all the patients.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.xnsj.2021.100090.

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