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IDEAS AND TECHNICAL INNOVATIONS

Resection of giant invasive sacral schwannoma using image-based customized osteotomy tools

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Abstract
Background Giant invasive sacral schwannomas are rare tumors. Surgical excision is the standard treatment and total resection is performed if feasible. Advances in three-dimensional (3D) imaging technology have facilitated treatment designs of complex surgical procedures.
Objective Our aim was to evaluate virtual surgical planning, computer-aided design (CAD), and manufacturing with 3D printing technology of the customized osteotomy guiding device in giant invasive sacral schwannoma resection.
Methods A digital 3D model of the sacrum, including the giant invasive sacral schwannoma, was rendered from patient computer tomography (CT) images. The surgeon chose excision margins of the tumor. Based on the virtual surgical planning, the customized guiding tool for osteotomy was designed and manufactured using the CAD and 3D printing.
Results We used the guiding block to successfully excise a giant sacral schwannoma using only a posterior approach to achieve gross total resection. No augmented spinal instrumentation was used to prevent iatrogenic spinal instability. Clinical symptoms resolved dramatically after operation. No spinal instability occurred during follow-up.
Conclusion With the assistance of an image-based customized osteotomy guiding device, we achieved both goals of tumor resection and bone preservation in giant sacral schwannoma resection. With thorough surgical planning, this technology can be applied to the complex surgical procedures easily and reliably.

Keywords Giant sacral schwannoma · Surgical excision · Computer-aided design · Guiding tool · CT imaging

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Introduction

Primary sacral tumors are rare and sacral schwannomas are even rarer. Only a few dozen cases are reported in the literature, most are case reports or series [1, 2]. The schwannoma is an encapsulated, slow-growing benign neoplasm, arising from a myelinated nerve sheath, the Schwann cell [1]. Although these nerve sheath tumors rarely arise within bone, they sometimes creep into adjacent vertebral body and extraspinal space. Sridhar et al. defined that giant invasive spinal schwannomas are masses that invade more than two vertebral levels, invade vertebral bodies, and by extending posteriorly, reach the myofascial regions [3].

Clinically, giant invasive sacral schwannomas are challenging given their invasive nature, complex anatomy, and difficulty in achieving safe margins. Currently, there is no established consensus regarding the best treatment strategies. The ideal treatment is complete removal of the tumor without neurovascular complications or spinal instability secondary to bone removal. This would relieve the neurovascular compression and regress symptoms [2, 4].

Based on the advances in three-dimensional (3D) imaging, virtual surgical planning (VSP) and computer-aided design (CAD) with 3D printing technology for the production of customized guiding device, surgeons, and bioengineers can work together to resolve the most complex surgical cases [5–7]. This study describes an image-based customized surgical guiding tool that supports surgeons working with giant invasive sacral schwannoma.

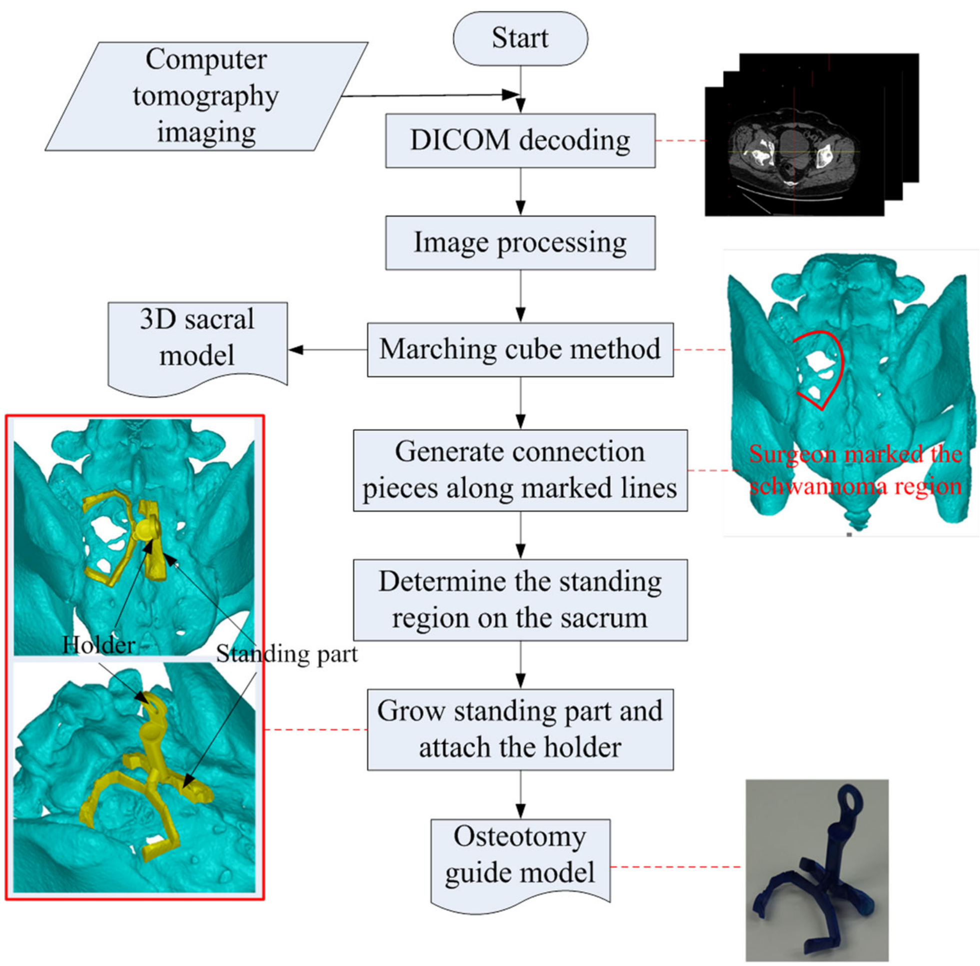
Materials and methods

Figure 1 illustrates the workflow for the design of the guiding tool. The DICOM CT data set was retrieved and decoded into general images for further image processing. Marching cube method was applied to reconstruct the sacrum model. Then, the surgeon marked the schwannoma region in our self-developed software. The osteotomy guide tool includes the holder, standing part, and resection C-ring for the schwannoma. The standing region was drawn on the dorsal surface on the median sacral crest of the 3D model. The standing part was grown from the marked region. A finger holder connects the standing part and the resection C-ring to form the osteotomy guiding device for further 3D printing.

Clinical presentation

A 23-years-old female patient presented to us with pain in the sacrococcygeal region and progressive voiding disturbance for months before the consultation. Image study revealed a giant invasive sacral tumor extending from S1 to S3. The tumor lesion not only changed the shape and composition of the bony construct, but also posed a difficult anatomical location for surgical resection. The 3D-reformatted computer tomography is shown in Fig. 2. She received a histological diagnosis using a computerized tomography-guided percutaneous needle biopsy. The

Fig. 1 Workflow for the design of the image-based customized osteotomy block



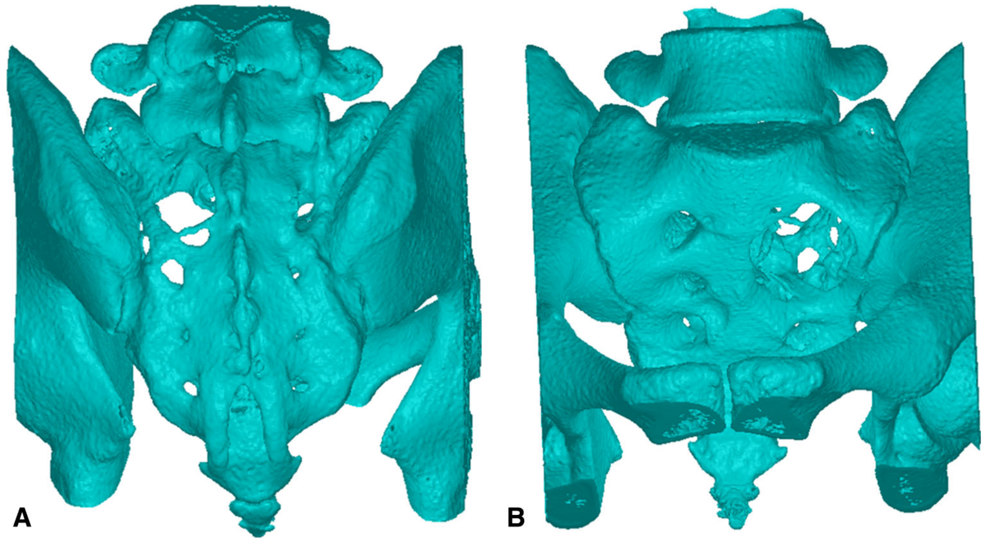


Fig. 2 3D reconstruction model of the sacrum. A giant invasive sacral tumor located from S1 to S3

pathology report confirmed the diagnosis of schwannoma. With thorough discussion, surgical resection was chosen as treatment.

Design principles of the osteotomy guiding device

The 0.625 mm computer tomography (CT) scan of the sacrum was obtained preoperatively. Based on the CT data, including the region of tumor, we reconstructed the 3D image model in our self-developed software for VSP. The image processing and marching cube methods were used for 3D image reconstruction [8]. The surgeon then determined the planned margins of resection in the software, which displayed the 3D model of the sacrum (Fig. 2). Since posterior surgical approach was decided, we created the base of the osteotomy guiding tools to stably fit on the dorsal surface on the median sacral crest. We then connected the base with a C-shape structure that marked and disclosed the resection area (Fig. 3). After thorough assessment and planning, the sacral model and customized guiding block were fabricated by 3D printing machine with acrylonitrile–butadiene–styrene (ABS) plastic material.

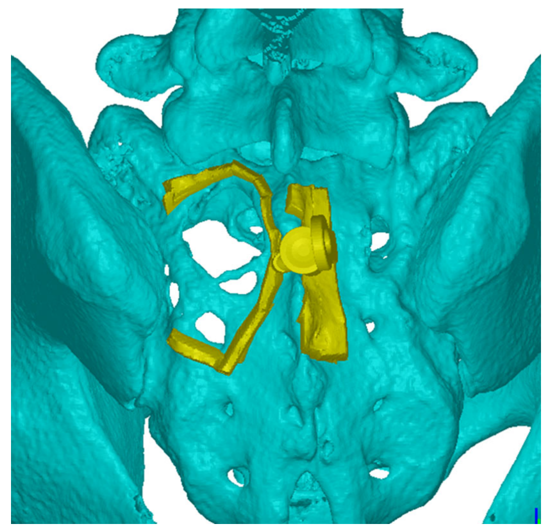


Fig. 3 C-ring of the guiding block defined the resection area

Resection margin planning

Planning the resection margin of the tumor is a critical step in the design workflow for the guiding tools. The physicians work hand-in-hand with engineering colleagues to exchange ideas and computer generated graphics. The engineer designed the connection pieces along the marked lines of the resection area defined by the surgeon. The specific needs of the patient and particular anatomic hazards are closely monitored. Comprehensive 3D surgical planning could be obtained by a user-friendly interface combining the CT images with sacral model in a 3D virtual environment. The digital data for the virtual surgical guide were rendered into an actual physical template using a stereolithography modeling technique.

Results

The posterior 'Y' incision was performed with the patient in the prone position (Fig. 4). The approach offered a good exposure of the sacrum, the dorsal sections of the iliac wings, and the surrounding soft tissues, while allowing exposure of the lumbar spine if necessary. As preoperative

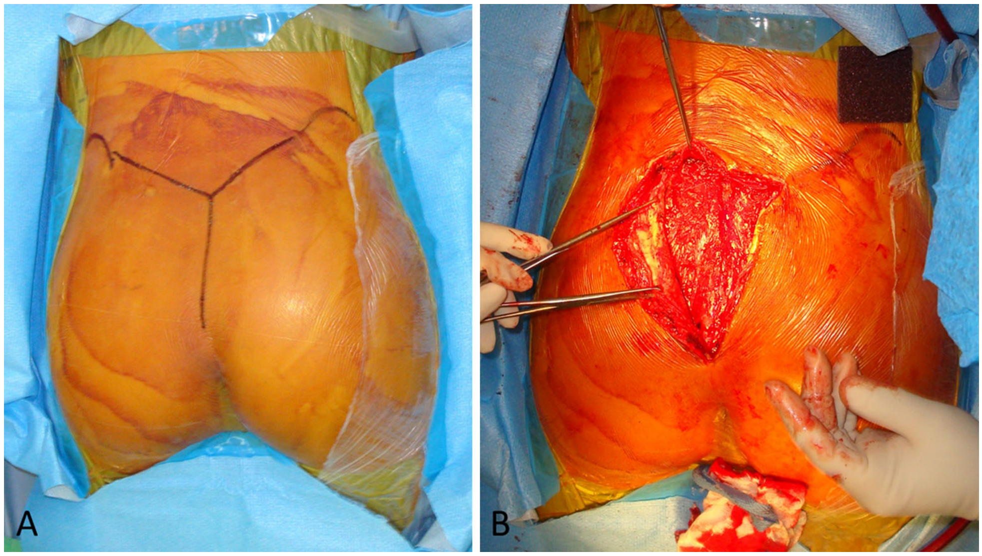


Fig. 4 Posterior 'Y' incision and flap design

planning in 3D-reformatted computer tomography and phantom bone model, this bone-model complex served as a dimension and shape reference to guide subsequent resection tasks (Fig. 5a). We localized the osteotomy margin of the tumor using the customized guiding block. To achieve accurate intraoperative localization, the standing platform of the customized guiding block was designed to rest closely on the median sacral crest in the contralateral side (Fig. 5b). The surface bony landmarks are critical in stabilizing the guiding device. The tumor was excised using a posterior approach, while the anterior aspect was removed with piece-by-piece manner, with care taken to ensure that the nerve and dura were preserved. The S1–S3 nerve roots were intact after resecting the tumor (Fig. 5c). No augmented spinal instrumentation was used to prevent iatrogenic spinal instability. Clinical symptoms resolved dramatically after operation. No spinal instability occurred during follow-up for 2 years. In this pilot project, we achieved both goals of tumor resection and bone preservation in giant sacral schwannoma resection.

Discussion

The use of VSP on the CAD software has gained popularity in clinical practice. The advent of 3D printing technology has further enhanced the quality and predictability of surgical procedures [9–11]. A variety of measurements, analysis, editing, and Boolean operations now provide surgeons with a clear picture of the human anatomy a vivid preview of postsurgical results. Based on the 3D models generated from CT scan, precise simulation of the resection area of a huge sacral schwannoma can be scheduled on computer preoperatively. With the supplement of modern technology, surgeons can perform surgeries with more confidence and accuracy.

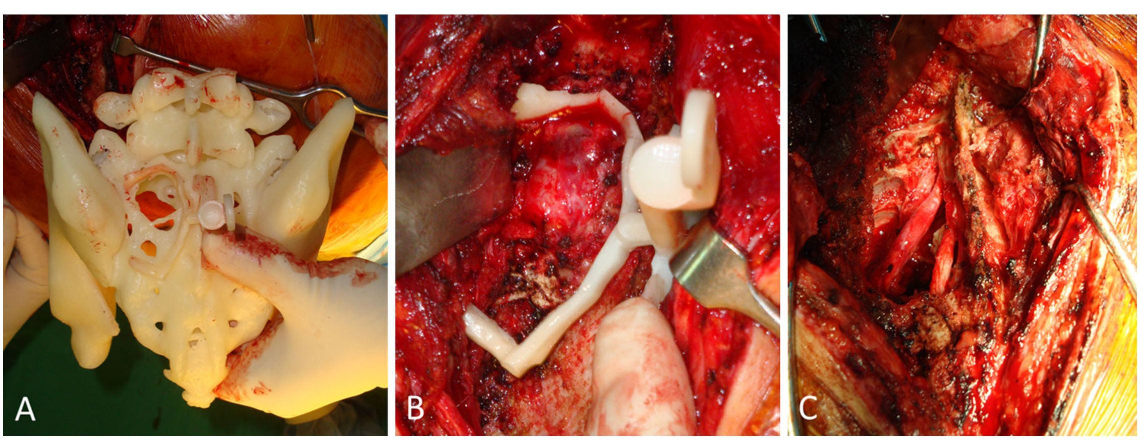


Fig. 5 **a** Bone-model complex. **b** Customized guiding block. **c** S1–S3 nerve roots were intact after tumor resection

Traditionally, surgeons preoperatively planned the resection margin according to 2D projective images or 2.5D stacked images and chose resection areas by direct vision intraoperatively. However, they usually needed to confirm the safe resection margin depending on the intraoperative frozen section [12]. It is difficult making these decisions intraoperatively. With this novel customized osteotomy guiding block, combined with intraoperative frozen section, we get a more accurate intraoperative localization of the resection margin of the tumor.

3D printing technology is widely used as an aid in the medical field, because it offers clear anatomical details with high-quality simulations of surgical procedures with the fabrication and adaptation of biomaterials [13]. Combining the interdisciplinary knowledge of surgeons and engineers, a custom-designed surgical guidance device, an osteotomy guiding block for sacral schwannoma was constructed with 3D printing technology. This technology has the advantage over the traditional surgical techniques of accuracy compared with the surgeon's choice of margins at the surgical site.

In our case, individual templates were customized on the basis of 3D reconstructions of the bone structures extracted from CT image data using self-developed software. A 3D printing machine was used to produce the shape of reference areas on the bone surface. The base of the surgical guide was "grown" on marked, non-destructible sacral surfaces. The holder was then added to the base of the surgical guide. To achieve accurate and stable intraoperative localization of the resection margin of the tumor, the platform of the customized guiding block was designed to rest closely on the median sacral crest. The holder is made crossing over the spinous process and the surgeon can hold it in place just above the vertebra. Our design required flexibility for varying environments (both small and stable) and complied with the above-described principles. The customized template can be sterilized and used intraoperatively. This technique reduced the time required for the operation and increased the accuracy of resection area.

Although this report shows the advantages of the guiding template, there are some limitations. Primarily, this preliminary study is only a case report with relatively short follow-up period. Second, the potential benefits of the surgery guided technique in improving surgical accuracy could not be proven by the control study without using the guiding template on the same patient. Nevertheless, further applications are expected to rely on the cooperation of surgeons and engineers.

Conclusion

In conclusion, this preliminary study demonstrates the use of VSP and 3D printing technology in the fabrication of an image-based customized surgical guiding device. This novel technique, stereolithographic model-guided osteotomy, will be the mainstay of the authors' approach to complex spinal tumor surgery. We believe that it has the potential to be a powerful tool for the most complex surgical cases.

Compliance with ethical standards

Conflict of interest The authors have no personal financial or institutional interests in any of the materials, or devices described in this article.

References

1. Togral G, Arikan M, Hasturk AE, Gungor S (2014) Incidentally diagnosed giant invasive sacral schwannoma. Its clinical features and surgical management without stability. Neurosciences (Riyadh) 19:224–228
2. Yu NH, Lee SE, Jahng TA, Chung CK (2012) Giant invasive spinal schwannoma: its clinical features and surgical management. Neurosurgery 71:58–66. doi:10.1227/NEU.0b013e31824f4f96
3. Sridhar K, Ramamurthi R, Vasudevan MC, Ramamurthi B (2001) Giant invasive spinal schwannomas: definition and surgical management. J Neurosurg 94:210–215
4. Ozdemir N, Bezircioglu H, Akar O (2010) Giant erosive spinal schwannomas: surgical management. Br J Neurosurg 24:526–531. doi:10.3109/02688697.2010.487129
5. Leon BR, Carrillo FJ, Gonzalez HM, Franco JL (1999) Mandibular reconstruction with the free vascularized fibular flap: utility of three-dimensional computerized tomography. J Reconstr Microsurg 15:91–97. doi:10.1055/s-2007-1000076 **(discussion 97–99)**
6. Su L, Dong Q, Zhang H, Zhou X, Chen Y, Hao X, Li X (2016) Clinical application of a three-dimensional imaging technique in infants and young children with complex liver tumors. Pediatr Surg Int 32:387–395. doi:10.1007/s00383-016-3864-7
7. Spetzger U, Frasca M, Konig SA (2016) Surgical planning, manufacturing and implantation of an individualized cervical fusion titanium cage using patient-specific data. Eur Spine J. doi:10.1007/s00586-016-4473-9
8. Lorensen WEC, Harvey E (1987) Marching cubes: a high resolution 3D surface construction algorithm. ACM Comput Graph 21:163–169. doi:10.1145/37402.37422
9. Noh K, Pae A, Lee JW, Kwon YD (2016) Fabricating a tooth- and implant-supported maxillary obturator for a patient after maxillectomy with computer-guided surgery and CAD/CAM technology: a clinical report. J Prosthet Dent 115:637–642. doi:10.1016/j.prosdent.2015.10.015
10. Farrell BB, Franco PB, Tucker MR (2014) Virtual surgical planning in orthognathic surgery. Oral Maxillofac Surg Clin N Am 26:459–473. doi:10.1016/j.coms.2014.08.011
11. Bobek S, Farrell B, Choi C, Farrell B, Weimer K, Tucker M (2015) Virtual surgical planning for orthognathic surgery using digital data transfer and an intraoral fiducial marker: the charlotte method. J Oral Maxillofac Surg 73:1143–1158. doi:10.1016/j.joms.2014.12.008
12. Bui MM, Smith P, Agresta SV, Cheong D, Letson GD (2008) Practical issues of intraoperative frozen section diagnosis of bone and soft tissue lesions. Cancer Control 15:7–12
13. Potamianos P, Amis AA, Forester AJ, McGurk M, Bircher M (1998) Rapid prototyping for orthopaedic surgery. Proc Inst Mech Eng H 212:383–393