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Resection of a sacral chordoma aided by neuronavigation: A case report

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ABSTRACT

Background: Chordomas are rare malignant neoplasms that are resistant to chemotherapy and conventional radiation therapy. The main factor that determines whether a localized tumor recurs is en-bloc resection with tumor-free margins. Intraoperative navigation is useful in this scenario, allowing for preoperative planning of the osteotomy.
Presentation of the case: A 60-year-old patient, who had been previously operated on for prostate cancer, presented with back pain after a minor accident. He was examined, and a sacral tumor that had been classified as positive for chordoma via biopsy was found. A neuronavigation-aided en bloc resection was carried out, with both S2 nerves, both sacroiliac joints and the coccyx all preserved.
Two years after the operation, the patient was asymptomatic, without tumor recurrence shown on MRI.
Conclusion: The use of intraoperative navigation is a useful tool for guiding osteotomies during bone tumor en-bloc resection.

1. Introduction

Chordomas are rare, malignant bone neoplasms that occur in less than 0.1 per 100,000 people per year [1]. Chordomas originate in parts of the notochord [2,3] and most often occur within the sacrum (50–60%), followed by the clivus (25–30%), the cervical region (10%) and, lastly, the thoracolumbar vertebrae (5%) [4,5].

Chordoma diagnosis sometimes takes time because the symptoms are usually nonspecific and long-term, which means that the tumor may have grown to a large size by the time it is diagnosed [6]. Most patients are diagnosed at Enneking's IB stage, and due to the proximity to neurovascular structures in the sacrum and pelvis, resection is a challenge for the lead surgeon [7,8].

Chordomas are normally resistant to chemotherapy or conventional radiation therapy [9], which is why surgical resection with tumor-free margins is the most frequently recommended treatment. Obtaining tumor-free margins means there is less likelihood of localized relapse [10,11] and a better survival rate [12].

Neuronavigation associated with intraoperative tomography allows for implants to be correctly placed, provides a multiplane visual of the anatomy during surgery and specifies the location of both surgical instruments and intrabone tumor lesions that are macroscopically invisible to the surgeon's naked eye. In the literature, there is evidence that navigation allows for preoperative planning to be followed, aimed at achieving tumor-free margins in bone tumor resection [13–15].

The aim of this study is to present and discuss the technical aspects of the surgical resection of a sacral tumor using neuronavigation.

2. Clinical case

A 60-year-old patient with a history of prostate cancer underwent a radical prostatectomy in 2016. At the beginning of 2017, he had a fall and went to the doctor with mild lumbosacral pain, as well as a sensation of anal sphincter disfunction.

A physical examination showed no superficial skin wounds in the lumbosacral or perianal region, and a neurological exam came back normal, without radicular irritation and showing an anal sphincter with a good tone.

Given his history of prostate cancer, he was sent for a SPECT CT, which showed an osteoblastic lesion of the sacrum at S3, so he was

Abbreviations: PET CT, Positron emission tomography-computed; MRI, Magnetic resonance imaging; SPECT CT, Single-photon emission computed tomography.

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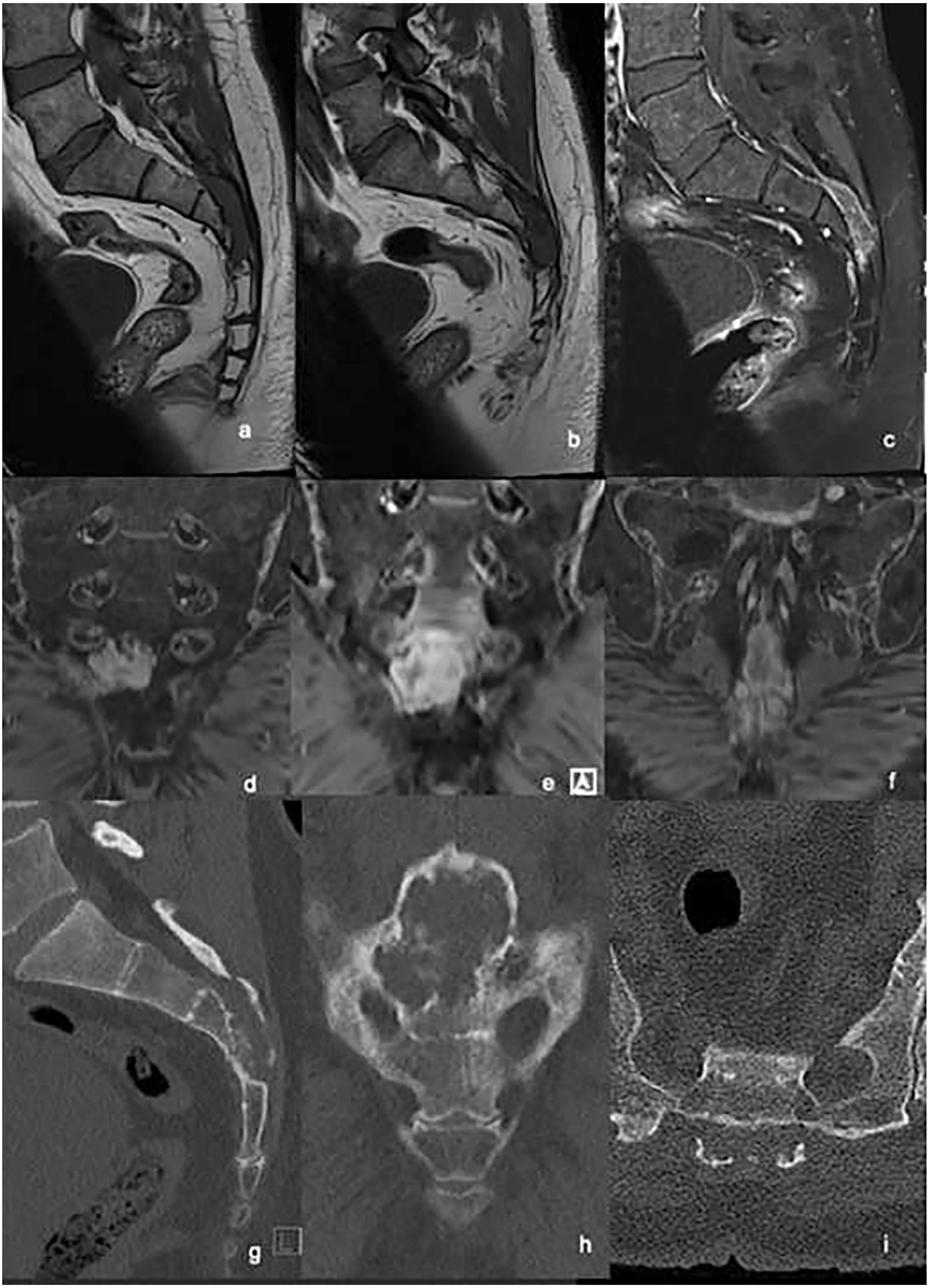

Fig. 1. MRI image showing the tumor from the bottom half of S2 to S4, with intraspinal proximity to S2 nerve roots and compromise of the right S3 root. (a, b) Sagittal and parasagittal T1 enhancement views. (c) Gadolinium enhancement view. (d,e,f) Coronal Gadolinium enhancement views from ventral to dorsal. (g,h,i) Sagittal, coronal, and axial CT scan images showing the tumor penetration to the dorsal cortex of the sacrum.

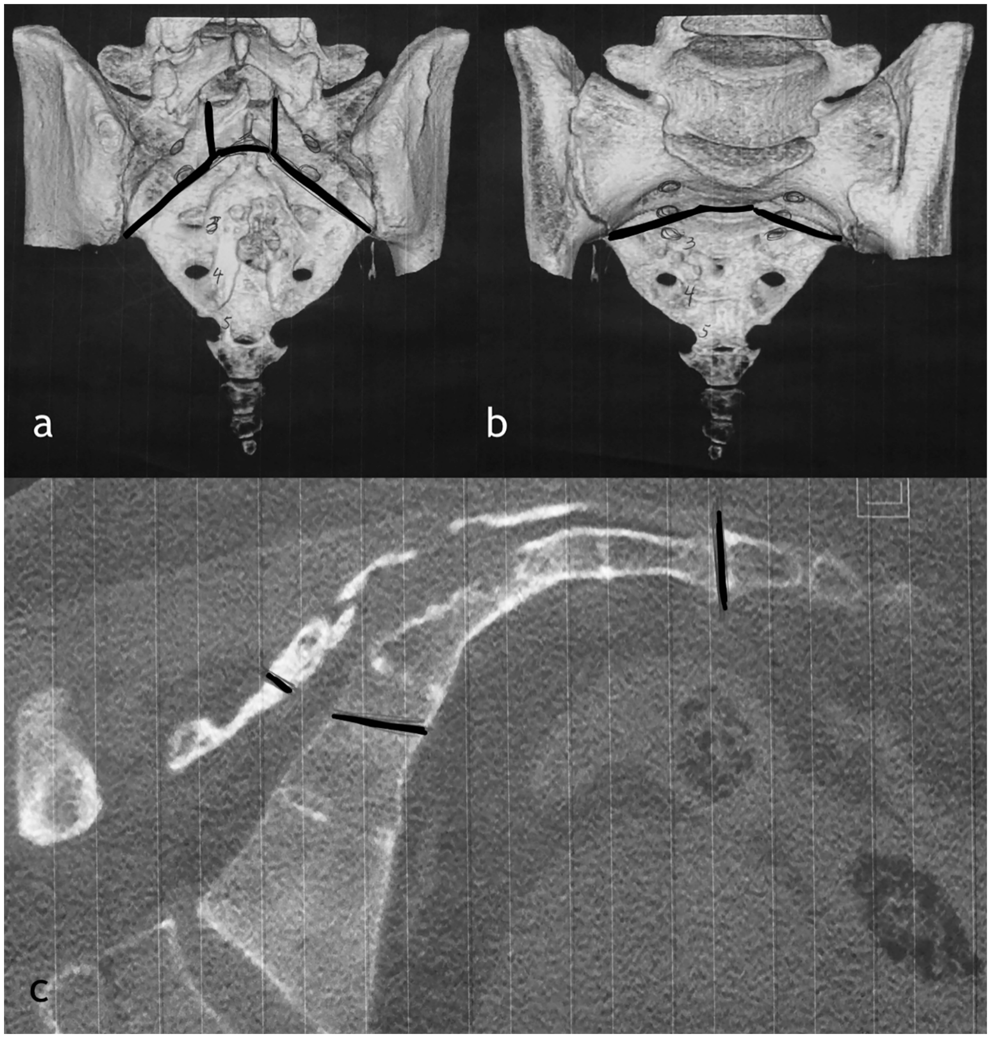

Fig. 2. Preoperative planification of the osteotomies: we did it drawing free hand lines over 3D reconstructions CT images. (a,b) posterior and anterior view of the sacrum showing the S1 laminectomy and the planned osteotomies through the foraminal S2 channel towards the inferior aspect of the sacroiliac joint. (c) sagittal reconstruction of the sacrum showing the cranial and caudal level of the osteotomies.

referred to the spinal surgery unit. In addition, a PET CT was carried out, which ruled out lesions in other locations.

The MRI showed a tumor extending from the bottom half of S2 to S4, affecting the S3 nerve roots and with intraspinal proximity to the right S2 nerve roots (Fig. 1).

A CT biopsy was carried out, and a histology study showed a unique low-grade chordoma at the GI, T2, N0, and M0 extra compartmental lesion stages.

Although the tumor was near the right S2 root and both S3 nerve roots were compromised, it was agreed upon with the patient that an en bloc resection would be scheduled, aided by neuronavigation for conserving the S2 nerve roots, both sacroiliac joints and the coccyx trying to achieve surgical free tumor margins.

We did a preoperative planification of the sacral osteotomies using 3D reconstructions CT images, we did not have a surgical planification software, so we did it drawing free hand lines over the preoperative CT images (Fig. 2). To ensure the intraoperative location of the osteotomy, we used navigation associated with an intraoperative tomography.

3. Surgical technique

Using a mid-line approach to resect the site of the biopsy, the posterior arch of L5 was exposed, to which the navigation guiding antenna was attached, and intraoperative tomography was taken. Next, a dorsal exposure of the sacrum, preserving the lumbosacral fascia like the dorsal margin of the tumor was performed. An S1 and S2 laminectomy was carried out, exposing, and tying the dural sac under the S2 nerve roots. Using the neuronavigation guide, a dorsal opening was made in the foraminal channel of both S2 nerve roots, and then a ventral osteotomy of the foraminal channel was carried out - matching the intraoperative location of the osteotome with the locations planned prior to the operation - from mid-S2 to the lower portion of the sacroiliac joints, the navigation system helped us to ensure to follow the pre-planned pathway and to avoid to violate the tumor. Later a transverse osteotomy of the sacrococcygeal union was performed by the same way, thus preserving the coccyx and its musculature. For the osteotomies, Kerrison rongeur, osteotomes and Gigli saws were used.

Making a blunt dissection, the sacrum was separated from the retro rectal space and was resected dorsally with the lumbosacral fascia to ensure a margin of soft tissue above the osteolysis dorsal tumor zone of the sacrum.

After the en-bloc resection, the plastic surgery team reconstructed the defect using a latero-medial gluteal advancement flap attached to the coccyx without having to use mesh (Fig. 3).

The surgery lasted five hours, and there was 400 cc. of intraoperative blood loss.

The anatomical-pathological result was a chordoma, and the borders of the resected piece of tissue were all tumor-free (Fig. 4).

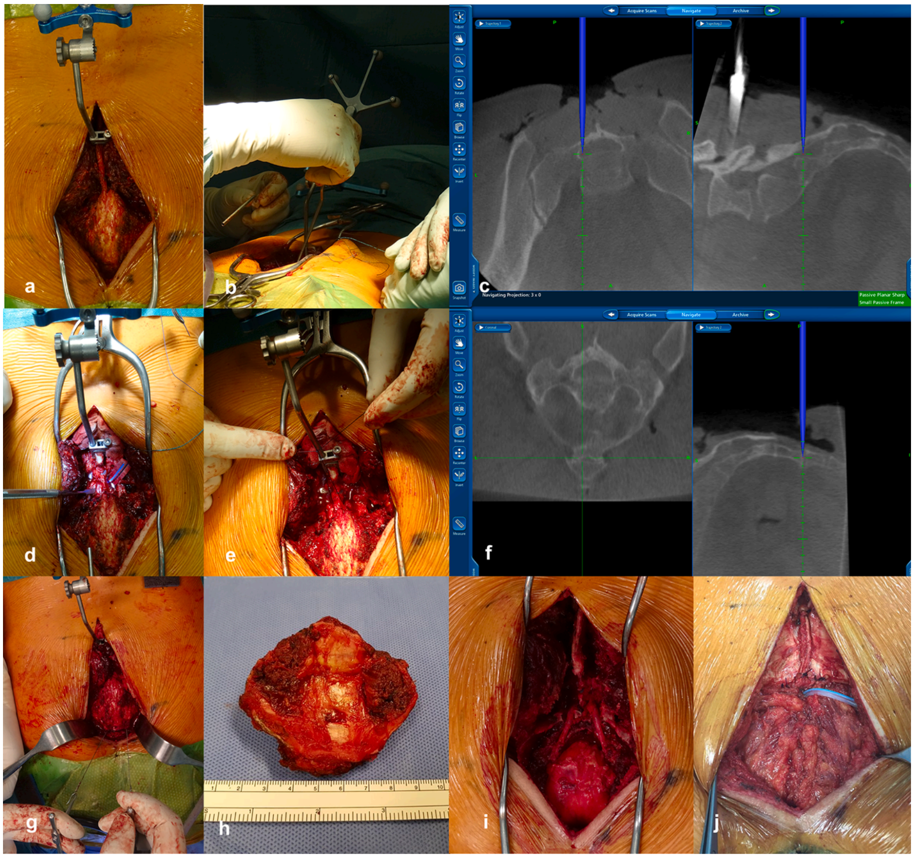

Fig. 3. Intraoperative pictures of the en bloc sacrum resection aided by neuro navigation: (a) Navigation antenna attached to posterior arch of L5. (b) intra-operative recognition of the sacrum anatomy with the navigated pinpoint. (c) Intraoperative navigation images showing the upper limit of the previously planned osteotomy. (d, e) exposure and ligation of the dural sac below S2 nerve roots. (f) Intraoperative navigation images showing the lower limit of osteotomy at the sacrococcygeal union. (g) Intraoperative section of the sacrococcygeal union with a Gigli saw. (h) Intraoperative piece of the sacrum. (i) The pictures show both S2 roots preserved and the rectum in the bottom part. (j) Reconstruction of the defect with two gluteal advancement flaps, attached to the remaining coccyx.

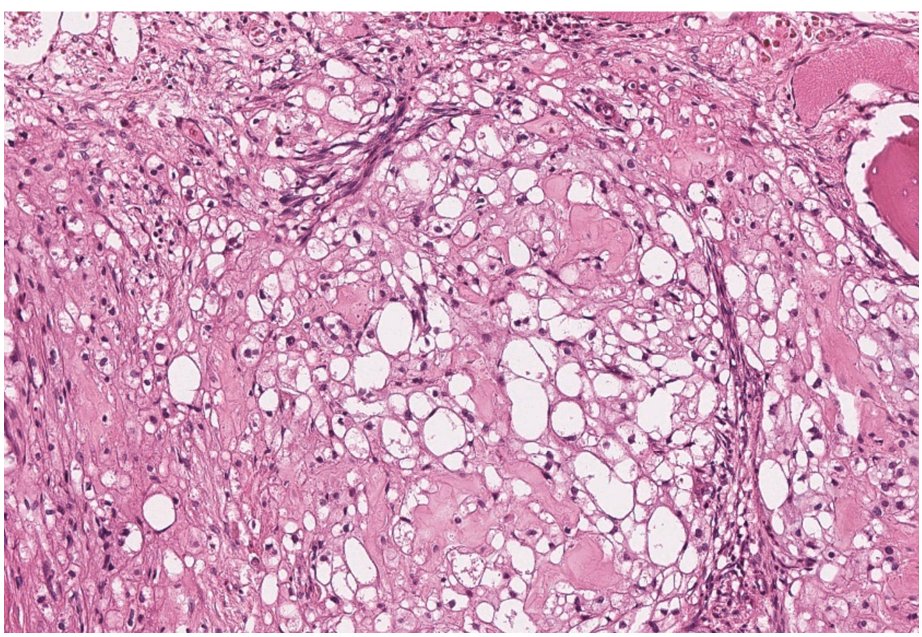

Fig. 4. 10x histological image, with bubbly appearance of Chordoma tumor cells (physaliferous cells).

The patient had no postoperative complications and maintained sphincter use and sexual function.

Two years after the operation, the patient was asymptomatic, without tumor recurrence shown on MRI or remotely. (Fig. 5)

4. Discussion

Chordoma resection is a challenging surgery since the proximity of the sacral nerves and pelvic organs makes resection with tumor-free margins and without compromising these structures [16]. Therefore, when an en-bloc resection is technically possible, any possible method or technique should be considered to ensure it is carried out properly

Fig. 5. Two years follow up after surgery: (a, b) Ap and lateral X rays. (c, d) MRI T1 and T2 weighted images of sagittal cuts, without tumor recurrence. We can see preservation of half of S2 body and the entire coccyx.

[12].

Our case involved a small tumor confined to extra compartmental S2-S4 with a violation of the sacral laminae and covered by muscle and lumbar fascia, which is why an en-bloc dorsal resection with wide margins and without the need for lumbo-pelvic reconstruction turned out to be the most logical approach.

Chordoma resection with wide borders is the procedure that most influences chordoma recurrence, as well as patient survival. Bergh published that patients who underwent surgical resections had a localized recurrence rate of 17%, while patients who underwent an intralesional or a marginal resection had a recurrence rate that increased to 81% [17].

This publication is consistent with other cases that highlight the usefulness of neuronavigation during the en bloc resection of sacral chordomas. Recently Konakondla et al published its case series showing similar results to our case, but with 5 months of follow up [18].

As far as the precision of neuronavigation is concerned Sternheim and colleagues [19] showed that navigated incisions were significantly more precise than non-navigated incisions, achieving a border of 5 mm between the target tumor mass and the plane of the planned incision in more than 95% of planned osteotomies. Factors that may affect this precision include preoperative planning using images obtained in the supine position, which changes to the prone position in surgery, leading to differences in the alignment of the vertebrae and discrepancies with what was planned in advance, as well as the use of navigation in mobile segments during the intraoperative period. In our case, we used intraoperative tomography in the final surgical position and, although we attached the antenna to the L5 bone - since we did not have a percutaneous method for anchoring it to the iliac crest - the work was carried out over the sacrum without inducing changes to its position regarding the pelvis or L5.

It is also well known that during conventional sacral chordoma resection, nerves are usually sacrificed with the aim of ensuring tumor-free margins. This sacrifice can cause important changes in sphincter and sexual function [7,8]. We know that vesical and intestinal function is mainly related to sacrificing the S2 nerve roots, while sexual disfunction is associated with S3 lesions [20,21]. In a systematic revision of 15 case studies involving 244 patients subjected to en-bloc resections for sacral chordomas, Zocalli and colleagues reported that, by preserving both S2 roots, there was a 39% chance of conserving vesical function, which decreased to 25% when only one S2 root was conserved [22].

In our patient, we decided to preserve both S2 nerve roots to allow better chance of sphincter and sexual function conservation, although the tumor extended halfway into this vertebral body, with the help of navigation, we were able to plan and to perform freeing up the S2 nerve roots, and we were able to do a trans-foraminal osteotomy from dorsal to ventral, corroborating in real time the position of the osteotome with a multiplanar view from the intraoperative tomogram, and being sure to follow the preplanned pathway, to avoid to violate the tumor margins, issue that is difficult to achieve using only conventional X-ray biplanar images.

On the other hand, given that the tumor was distant from the sacrococcygeal union, we decided to preserve the coccyx with all its musculature, which benefited the intraoperative procedure since the plastic surgeons did not need to use a restraining mesh for closure and could attach the gluteal advancement skin flaps to the coccyx.

Our patient ended up with annal sphincter a sexual function conservation, better than expected. The facts that may be related to a sphincter training after his cancer prostatectomy, a probable predominant S2 pudendal nerve innervation and the coccygeal muscle conservation could help in the annal function.

We believe that the use of intraoperative navigation is a tool that optimizes the surgeon's vision and provides the opportunity to visualize the tumor's anatomical location and that of the surgical instruments in different tomographic planes in real time. This technique can help maintain sufficient distance from the tumor, preventing both the tumor from opening up and the subsequent contamination of the surgical site, thus helping to more easily achieve the aim of tumor-free margins and fewer recurrences.

Undoubtedly, intraoperative navigation systems are expensive to implement, but their cost efficiency for implants has already been suggested. Based on our limited experience, if this technology is available, we recommend its use for planning the resection of a sacral tumor or other bone-located tumors since navigation can increase the probability of an en-bloc resection and ultimately the patient's survival rate.

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.

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