



## Technical Note

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# Sacral Nerves Reconstruction After Surgical Resection of a Large Sacral Chordoma Restores the Urinary and Sexual Function and the Anal Continence

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**Objective:** Chordomas are slow-growing tumors, with a high tendency to local relapse. *En bloc* resection is related to the most favorable outcome in terms of survival but is frequently associated with permanent neurological deficits involving sphincters and sexual functions. In the present article, we describe an innovative technique of *en bloc* resection followed by reconstruction of the sacral nerves with nerve grafts.

**Methods:** The chordoma was excised through a posterior approach after dividing the proximal and distal sacral nerves using the established technique. After that, a microsurgical S2-S3-S4 nerve reconstruction was performed connecting the proximal and distal stumps with sural nerve grafts withdrawn from both lower limbs.

**Results:** Immediately after surgery, the patient experienced complete impairment of sexual function and sphincters with urinary and fecal incontinence. After 6 months, there was a progressive recovery of sexual function and sphincter control. One year after the operation, the patient achieved an adequate sexual life (erection and ejaculation) and complete control of the bladder and anal sphincter.

**Conclusion:** Reconstruction of nerves sacrificed during sacral tumor removal has been shown to be effective in restoring sphincter and sexual function and is a promising technique that may significantly improve patients' quality of life.

**Keywords:** Chordoma, Nerve reconstruction, Fecal incontinence, Complications



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## INTRODUCTION

Chordomas are rare and locally aggressive tumors arising from remnants of notochord.<sup>1,2</sup> They were first described in the literature in 1857<sup>3</sup> and account for 1%–4% of all primary bone tumors. Despite the small growth rate, the incidence of recurrences ranges from 66% to 75%,<sup>4-6</sup> while the occurrence of metastasis<sup>7,8</sup> to the bone, lungs, and liver<sup>9</sup> is very low.

Chordomas typically occur in adults between the ages of 40

and 70 and affect males twice as often as females.<sup>8,10</sup> They are classified into 4 groups based on anatomical location: skull base (25%–40%), cervical spine (10%), thoracolumbar spine (5%), and sacrum (50% to 60%).<sup>8,11,12</sup> Due to the slow growth of the tumor mass, pain is the most common presenting symptom for sacral chordomas (SC),<sup>13-15</sup> but patients may also experience neurologic symptoms when the lesion involves the nerve roots, characterized by changes in bowel and bladder function, incontinence, sexual and walking impairment.

Treatment options for SCs include surgical resection, radiotherapy, and chemotherapy.<sup>16</sup>

Whenever feasible, surgical *en bloc* resection of the tumor, extended to surrounding normal tissues, is considered the optimal treatment option in the first instance.<sup>11,17-19</sup> Indeed, total resection is associated with longer relapse-free survival rates.<sup>11,14,20</sup>

Although radical excision correlates with better survival outcomes, patients undergoing SC surgery face a high morbidity rate.<sup>14</sup> Major complications include: blood vessel injuries (hypogastric and iliac vessels), visceral injuries (bowel and urinary tract), neurological deficits (neurogenic bowel and bladder, sciatic nerve or lumbosacral plexus injury<sup>21</sup>), instrument failure (related to spinopelvic instability<sup>10,11</sup>), wound infections and complications such as wound dehiscence and surgical site infection.<sup>22</sup>

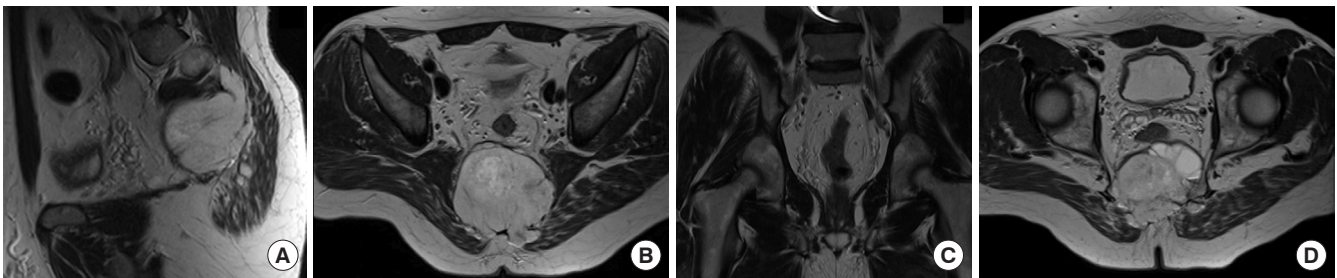
The purpose of this paper is to describe the technique used for *en bloc* removal of a large SC involving all sacral nerve roots below S1 on both sides, where the nerves divided and excised

along with the tumor mass were reconstructed in the same procedure using nerve grafts.

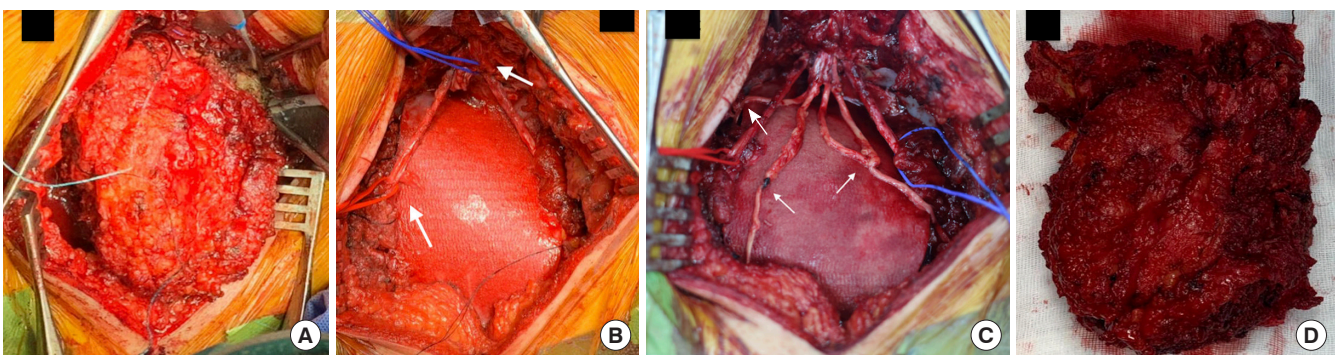
## MATERIALS AND METHODS

### 1. Case Description

A 54-year-old man with no medical history has come to medical attention for low back pain and coccydynia associated with lower limb discomfort and numbness, which has gradually increased over the last months. For these reasons, he was advised to take an x-ray of the lumbosacral spine, which showed bone erosion of multiple sacral metamers. Enhanced gadolinium magnetic resonance imaging (MRI) revealed a lesion extending from the inferior margin of S1 to the coccyx (size: 9 cm × 8 cm × 6 cm) with bilateral infiltration of the gluteus maximum muscle and iliac wings. The lesion appeared as an irregularly shaped lobulated mass with heterogeneous intensity and enhancement (Fig. 1). On inspection, no masses were visible and the patient



**Fig. 1.** Magnetic resonance imaging (MRI) revealed a lesion extending from the inferior margin of S1 to the coccyx (size: 9 cm × 8 cm × 6 cm) with bilateral infiltration of the gluteus maximum and iliac wings. The lesion appeared as an irregularly shaped and lobulated mass with heterogeneous intensity and enhancement: sagittal T2 view (A), coronal T2 view (B), axial T2 views (C, D).



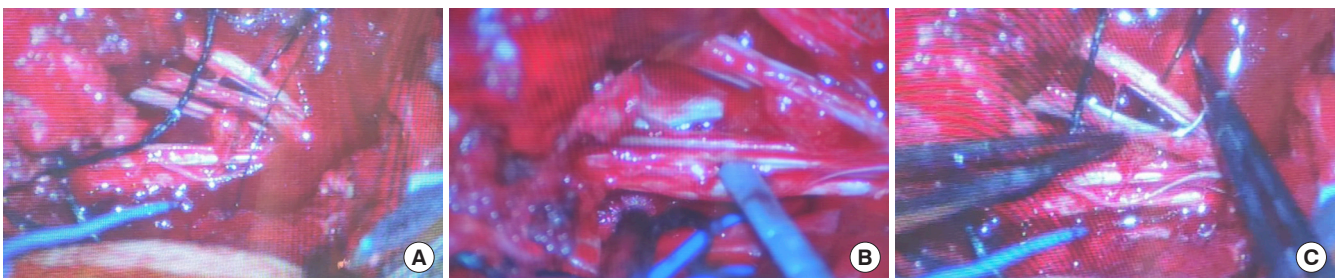
**Fig. 2.** Intraoperative images during sacral chordoma removal. (A) Initially, a bilateral dissection was performed within the gluteal muscles, until reaching the healthy bone plane, lateral to the sacro-ileal joints. (B) Sutures were applied to the perineurium of the extradural portion of the roots before they were cut (white arrows). (C) Bilateral sural nerve withdrawal was performed and 4 reconstruction grafts with perineurium microsutures were made between the proximal and distal nerve stumps, connecting S2 to a distal cranial end and S3-4 along with one end of the distal nerve caudal on each side (white arrows). (D) The bulky mass, containing the sacrum and sacral nerves, was removed *en bloc*.

was neurologically intact. Computed tomography (CT) with 3-dimensional reconstruction revealed erosion of the sacrum and infiltration of the lower segments of the iliac bones, while the upper portions of the sacroiliac joints were free from infiltration. After reviewing the images and discussing the case, we planned the *en bloc* resection through a single-stage posterior approach. The patient signed a specific and extended informed consent, following the opinion of the institutional review board.

## 2. Surgical Technique

The patient was placed in the prone jackknife position, under general anesthesia. Lower limbs muscles and sphincter functions were under continuous intraoperative neurophysiological monitoring. A posterior midline incision was made from the L4 spinous process down to the tip of the coccyx. A lateral dissection was initially carried out within the gluteal muscles, until reaching the healthy bone plane, laterally to the sacro-ileal joints (Fig. 2A). After performing a L5-S1-S2 laminectomy, the distal end of the dural sac, the roots of the cauda equina in their extradural portion distally to the dorsal root ganglion, and the filum terminale externum were identified with the aid of the operating microscope. Electrical stimulation was used as a method of mapping. The cranial extension of the tumor mass was assessed, in order to spare the nerve roots not affected by the tumor (in this case L5 and S1 on both sides). The roots below S2 were encased by the tumor mass and it was not possible to spare them and remove the lesion radically. S1 osteotomy was performed in the ventral direction, above the cranial portion of the lesion. Next, a bilateral osteotomy was performed, extending lateral to the ileum in the lower third of the sacroiliac joints. Osteotomies were performed with a high-speed drill, ultrasonic bone aspirator and completed with bone rongeurs. At this time the piriformis muscles and the sacro-spinous, sacro-tuberous and sacro-ileal ligaments were cut. The cranial portion of the sacrum was then mobilized and progressively dislocated in the

cranio-caudal direction, while the dural sac was under direct vision and, with the fulcrum of rotation at the level of the coccyx, still bound by the ligament structures. In order to remove the sacrum while preserving S1 roots, the S1 foramina were unroofed and the S1 roots were displaced anteriorly after dividing the small posterior sensitive branches outgoing from the S1 posterior sacral foramina. Next, landmark sutures were applied to the epineurium of the extradural portion of the S2-S3-S4 roots before they were cut. (Fig. 2B). We proceeded with the cranio-caudal dislocation of the sacrum and the tumor mass, which was bluntly dissected from the rectum and from the visceral and pelvic vascular structures with the aid of a gauze. The tumor-sacrum complex could be mobilized in the cranial portion but was still constrained inferiorly by the anococcygeal ligament and by the distal portion of the sacral roots, in the portion where they emerged from the tumor. The cranio-caudal dislocation of the sacrum continued, including the tumor mass, which was bluntly dissected from the rectum and pelvic visceral and vascular structures with a gauze. At this time the tumor-sacrum complex could be mobilized in the cranial portion but was still constrained inferiorly by the anococcygeal ligament and by the distal portion of the sacral roots, in the tract where they emerged from the tumor. It is precisely the mobilization of the mass that has allowed the identification, both visual and tactile, of the nerve trunks distal (caudal) to the lesion, before the point where they merge to form the sacral plexus and pudendal nerve. Landmark sutures were then applied to the epineurium approximately 2 cm distal from the tumor and these nerves and ligament structures were divided, allowing for *en bloc* removal of the mass (Fig. 2C, D). The application of stitches on the epineurium has the purpose of providing landmarks and avoiding losing the distal stumps of the sectioned nerves. Immediately after sectioning the nerves, there was a rapid decline in recordable potentials. Several histological samples taken from the margins of the lesions were sent for intraoperative his-



**Fig. 3.** Intraoperative microsurgical view. (A) Exposure of the nerve roots proximal to the tumor (mass to the left) under magnification. (B) Neurophysiological mapping with neuro-stimulator. (C) Epineural microsuture.



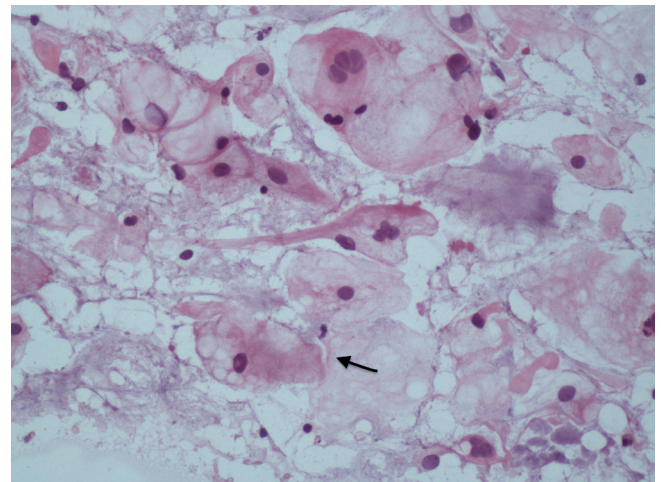
tological examinations, which revealed the negativity of the margins and prerectal fat. A resorbable prosthesis (GoreBioA) was placed on the rectal plane in order to prevent posterior visceral herniation (Fig. 3).

Bilateral sural nerve withdrawal was performed and 4 reconstruction grafts with epineurium microsutures were performed between the proximal and distal portions of the nerves, connecting S2 to the most cranial distal nerve stump and S3–4 together with the most caudal distal nerve stump on each side, before they merge to form the sacral plexus (Fig. 4A, B). Fibrin glue was applied to reinforce neurorrhaphy. The cavity was washed with normal saline, bleeding was controlled using gauze and hemostatic agents, a drainage tube was placed, and the soft tissues were closed with multilayered direct sutures. It was not deemed necessary to proceed with stabilization and fusion. The reconstruction of the muscular, subcutaneous and cutaneous planes were performed in a standard way.

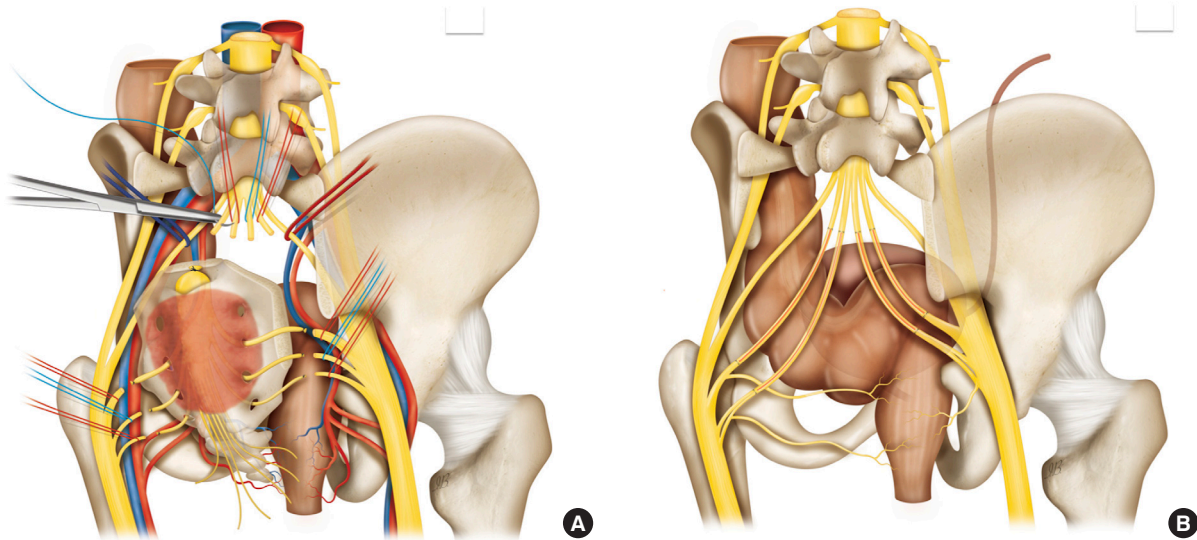
## RESULTS

Histological examination of the tumor specimens, measuring 14 cm × 8 cm × 7 cm, revealed typical physaliphorous cells with intracytoplasmic mucin-like substances, large nuclei, deep staining, and an irregular matrix that resembled stroma, cartilage, and mucus. Immunohistochemistry revealed that the specimens were positive for cytokeratin, epithelial membrane antigen, Ki-67, S100, and vimentin. Based on the findings from the imag-

ing and the pathological results, the final diagnosis was sacral chordoma (Fig. 5). After surgery, there was an immediate onset of complete sphincter deficiency, as expected. Instrumental examinations, electromyography (EMG) of the anal sphincter and rectal manometry, showed complete denervation with fibrillation potentials and the absence of effective sphincter tone. The patient maintained bladder catheterization and had uncontrolled fecal evacuation. Sexual impotence was also associated. The wound presented dehiscence, which was resolved with repeated



**Fig. 5.** Histological examination of the specimen revealed typical vacuolated physaliphorous cells (arrow) with intracytoplasmic mucin-like substances, large nuclei, deep staining, and an irregular stromal, cartilage, and mucinous matrix.



**Fig. 4.** Schematic drawing of the surgical steps. (A) Sacral chordoma enclosing multiple sacral roots. The recognition of the nerve roots and their marking with sutures in the healthy portions proximal and distal to the lesion are necessary in anticipation of the subsequent nerve grafting procedure. (B) Interposition grafts were performed in order to allow the subsequent reinnervation of the pudendal plexus to restore anal and genitourinary sphincter functions.

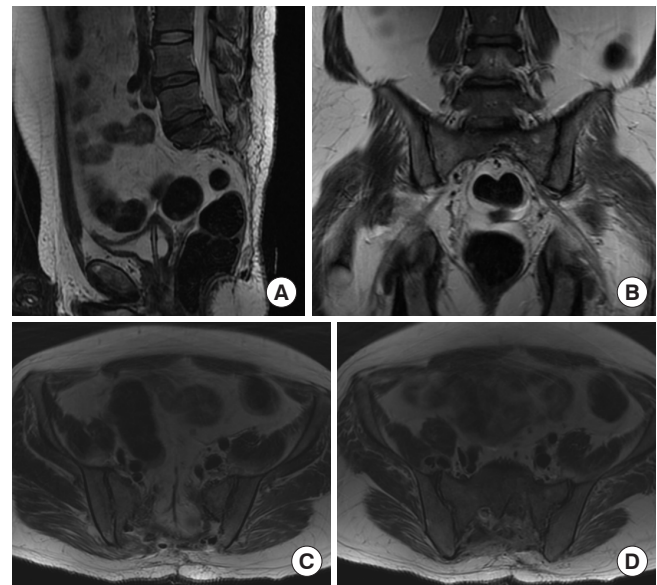


**Fig. 6.** Anal sphincter electromyography. (A) The postoperative exam 3 weeks after surgery shows acute muscle denervation with fibrillation potentials. (B) The electromyographic pattern 1 year after surgery shows normal interference activity.

dressings, and the patient was finally discharged on the 27th postoperative day. Given the radical removal of the lesion, the radiation oncologist, also taking into account the patient's wishes, did not indicate an adjuvant treatment and the patient was referred to a neurourological rehabilitation center where he began to perform intermittent catheterization and had a postvoiding residue of about 250 mL. In the following months, the patient presented a progressive improvement of the sphincter function, suggesting reinnervation through the grafts. At the 3-month follow-up, the discomfort in the lower limbs began to improve. At 6 months after surgery, the patient began to feel the sensation of bladder filling and the postvoiding residue was progressively reduced. He began to empty the bladder spontaneously without the need for catheterization. At 1 year of follow-up, the ultrasound showed an almost normal postvoiding residue of 30 mL. Sensitivity in the sellar and genital region and sexual function were also progressively recovered and the patient was able to have an erection and ejaculation. Fecal control was also progressively achieved and the patient was weaned from the diaper at 8 months after surgery. CT, MRI, and a clinical examination revealed no signs of tumor recurrence. Anal manometry showed anal sphincter tone recovery and EMG showed reinnervation of the anal sphincters (Fig. 6). At 1 year of follow-up, the patient was able to lead an almost normal life and the MRI showed no recurrence of the disease (Fig. 7).

## DISCUSSION

SC surgery must take into account both oncological and functional outcomes. The first relates to tumor residue after surgical resection and predicts local control of primary sacral disease and relapse-free survival.<sup>23</sup> The latter includes several components: spinal stability, deambulation, sphincter control, and sexual function, and affects the health-related quality of life. The *en bloc* excision is considered the best surgical therapeutic choice



**Fig. 7.** Magnetic resonance imaging (MRI) performed 1 year after surgery showed no signs of tumor recurrence. (A) Enhanced sagittal T1 MRI, (B) sagittal T2, (C, D) axial enhanced T1.

from an oncological perspective.<sup>11,24</sup>

SCs can often grow long before becoming symptomatic and, when the lesion involves the superior sacral nerve roots and complete resection is planned, serious complications such as neurological bladder and incontinence are to be expected.<sup>25</sup> The burden of the postsurgical complications of sacrococcygeal chordoma removal is so massive that sacral amputation is advisable only when it fits the radical excision criteria and allows for significant prognostic benefits.<sup>26-28</sup> Patients facing neurological bladder and fecal incontinence after sacrectomy require long-term enema, diapers, and urinary catheterization.<sup>29,30</sup> Sexual impotence accompanies the clinical picture and, when the S1 root is involved, walking is also impaired. Intralesional tumor removal can be offered to patients who do not want to deal with these complications. However, intralesional excision is associated with a high local recurrence rate and a poor prognosis.<sup>24,15</sup>

In the available literature, most of the articles focus on the choice of the approach to the tumor (anterior, posterior, or combined),<sup>31,32</sup> to the reconstruction of the large postsurgical soft tissue gap and to the postprocedural spinopelvic stability and related instrumented reconstruction techniques.<sup>33,34</sup> Despite the great attention paid to this type of technical aspect, less effort has been devoted to rescuing or restoring neurological function. Some of them are the technical variants to the original total sacrectomy, which have been proposed in order to reduce

morbidity and improve the functional results, without neglecting the total removal. These techniques are based on previous studies showing that unilateral sparing of sacral nerve roots usually results in the preservation of sphincter function, although there is no firm rule in predicting postoperative function.<sup>25</sup> Some data suggest that sparing of bilateral S3 sacral roots is not associated with postoperative disturbances of sphincter function<sup>35</sup> while other observations show that sparing at least one S3 sacral nerve root is associated with maintaining bladder function in most patients.<sup>25,31</sup> According to Nakai et al.,<sup>36</sup> on the other hand, the sacrifice of a sacral root S3 results in bladder or bowel dysfunction, albeit incomplete. Consequently, these surgical models were developed to adapt the extent of excision/debulking to the extent of the disease with the aim of sparing as much as possible the sacral roots and nerves.<sup>37</sup> However, since the negative margin excision is a cornerstone of this surgery, the nerve-sparing procedure is selectively dedicated to cases where there is no infiltration of neural structures.

Larger SCs, extending up to the S2 level, are characterized by bilateral root infiltration and their *en bloc* removal is associated with a high rate of permanent neurological deficits.

To address this problem, we planned a total removal of the lesion and the roots involved, followed by the reconstruction of the nerves by interposing autologous grafts. Nerve reconstruction by grafting is a long-standing acquisition of surgery, but the application of this technique in this area after excision of a sacral neoplasm has never been previously reported. This type of technique had so far only been hypothesized or tried at an experimental level. It has been shown in a canine model that the section of the sacral nerves that innervate the bladder, followed by end-to-end reconstruction, leads to a recovery of bladder function.<sup>33</sup> Other studies have investigated surgical strategies to restore the function of a denervated bladder after neurotrauma by transferring new axonal sources, but the techniques described and reported in the literature are only theorized or tested on animal models and never used in the clinical setting.<sup>33,38</sup> In this case, we have shown that this reconstructive surgery and recovery of function is also possible in the clinical setting. Since the tumor envelops the nerves, the removal of the lesion causes the formation of a large nerve gap so end-to-end repair is not feasible and the use of graft is mandatory. Regarding the nerve grafts, one of the issues that must be considered includes whether it is long enough to ensure a tension-free anastomosis.<sup>39</sup> The potential donor nerves for autografting are generally limited to cutaneous nerves of the extremities, and the sural nerve, in the lower limbs, is considered to be the standard for nerve grafts.<sup>40</sup>

Despite the biological superiority of autografts, their withdrawal adds morbidity to the intervention and some alternatives can be considered such as cadaveric nerve allografts and nerve guidance conduits.<sup>41-43</sup>

The use of neurophysiological monitoring can be helpful for mapping. However, we believe that it is not essential because the anatomical mapping is very reliable and, after the nerve section, the possibility of recording nervous activity is immediately lost. During the follow-up period, there was a progressive recovery of the function of the anal and bladder sphincters and of sexual function. These data, confirmed by instrumental tests, suggest reinnervation through the nerve grafts. Postoperative radiation therapy was not performed. The use of postoperative radiotherapy is generally recommended but, in the case of radical removal, the guidelines do not consider it mandatory.<sup>23,44</sup>

Despite the advantages described, this technique has some limitations. First of all, the technical difficulty that requires skills in different fields (oncological surgery, orthopedic surgery, neurosurgery, peripheral nervous system surgery). Furthermore, there is a difficulty in standardizing the technique due to the high complexity and variability of SCs.

However, after our preliminary experience, we believe that nerve reconstruction in SC surgery is a promising technique, capable of having a favorable impact on patients' quality of life without compromising oncological excision. Long-term monitoring will be needed to identify any recurrence or metastasis. Further studies are planned, both on cadaveric specimens and in clinical settings, in order to validate and disseminate this technique.

## CONCLUSION

The reconstruction of the nerves after the radical removal of a huge SC made it possible to recover the sphincter and sexual function that were impaired after the surgery. To our knowledge, this is the first report of a reconstructive technique applied in this context.

## NOTES

**Conflict of Interest:** The authors have nothing to disclose.

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**Author Contribution:** Conceptualization: LB; Data curation: DA, EM; Methodology: MP; Project administration: LB; Writ-



ing - original draft: LB, DA, MP, MV, EM; Writing - review & editing: LB, VDN, LD, MM, AS.

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