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Surgical treatment of giant benign sacral neurogenic tumors using the posterior-only approach

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ABSTRACT

Objective: Few studies have reported on the effects of surgery on giant benign sacral neurogenic tumors since these tumors represent a major challenge for surgical oncologists. In the present study, we retrospectively reviewed our experiences in performing resections of giant benign sacral neurogenic tumors using the posterior-only approach via "fishing" method, and assessed the outcomes of patients after this procedure.

Patients and methods: We retrospectively reviewed and analyzed the records of 155 consecutive patients with giant benign sacral neurogenic tumors who underwent resection by a posterior-only approach in our center from Feb 2008 to Feb 2018.

Results: Operations lasted for a mean of 183 ± 37 min (range, 125–230 min). The mean estimated blood loss during the surgery was 1098 ± 889 ml (range, 600–4100 ml). The mean follow-up time was 65 ± 39 months (range, 12–118 months). By the last follow-up, the local recurrence rate was 7.1% (11/155), the combined overall symptom resolution and remission rate was 91.1% (123/135), and the total complication rate was 17.4% (27/155).

Conclusion: The posterior-only approach is safe and effective in patients with giant benign sacral neurogenic tumors.

1. Introduction

Neurofibromas and schwannomas are benign neurogenic tumors arising from sacral nerves. These tumors, when entering the presacral space, may grow into huge masses. Although not necessary for symptomatic benign neurogenic tumors, treatment is needed for those presenting symptoms from compressing the rectum, bladder, ureter, and/or adjacent nerves. The current treatment options include surgery, interventional perfusion, radiofrequency ablation, endovascular embolization, and computed tomography (CT)-guided iodine-125 (125I) seed implantation [1,2]. Surgical resection remains the optimal treatment option for patients with giant benign sacral neurogenic tumors [3], yet it presents a major challenge for surgical oncologists because of the complex anatomical structure, the difficulty in preserving the function of nerve roots upon removing the tumor, and the possibility of massive bleeding during the operation. For many years, surgeons have attempted to develop solutions to both safely remove the tumor with minimal blood loss and preserve the function of the affected nerve roots as much as possible. Due to the relative low incidence of sacral neurogenic tumors, few studies have comprehensively reviewed the effects of surgery on these tumors. In 2009, we reported our experiences on surgical treatment of 48 cases of sacral neurogenic tumors [2]. Here, we retrospectively reviewed the outcomes of surgery using the posterior-only approach on 155 patients with giant benign sacral neurogenic tumors.

2. Patients and methods

2.1. Patients

This study was approved by the Institutional Review Board of our hospital. We retrospectively reviewed the records of 165 consecutive patients who underwent surgical resection of the giant benign sacral neurogenic tumor using the posterior-only approach in our institute from Feb 2008 to Feb 2018. Ten patients who were lost during the follow-up were excluded from this study. The remaining 155 patients included 76 males and 79 females with a mean age of 46 years (range, 18–67 years). The pathologic diagnosis was neurofibroma in 94 patients and schwannoma in 61 (Table 1). The major clinical manifestations included pain in a lower limb or sacrococcygeal region (n = 90), bladder and bowel dysfunction (n = 31), neurologic deficit (n = 14), and palpable painless mass (n = 13). Besides, seven patients were identified during routine physical examination (Table 2). Of the 155 patients, 135 received primary treatment and the remaining 20 were referred from other hospitals because of tumor recurrence. The mean

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Table 1
Clinicopathological characteristics of patients in this study.

Clinicopathological Characteristics	
Gender	
Male	76
Female	79
Age in years (range)	46 (18-67)
Tumor size (diameter, cm)	11.1 ± 3.6
Pathologic diagnosis	
Schwannoma	61
Neurofibroma	94

diameter of the tumors was 11.1 ± 3.6 cm (range, 5–21 cm). The diagnosis of benign neurogenic tumors was established on characteristic imaging features (displaying a homogenous, well-defined tumor mass on CT images) in 143 patients and through CT-guided biopsy in 12 patients (displaying non-homogenous signals on CT images), and all confirmed by pathologic examinations during the surgery.

Complications, including surgery-related complications and mechanical failures, were determined at the final follow-up. A major complication was defined as one that necessitated additional surgical procedures. A minor complication was defined as a problem that required nonoperative management.

2.2. Surgical procedures

Surgical indications for resection of benign sacral neurogenic tumors using a posterior-only approach in our series were: (1) the tumor was diagnosed as a benign mass, and (2) the upper level of the tumor was below the S1 level.

Operation procedure: Before the operation, intra-arterial embolization was performed in the Radiation Department via femoral access to selectively embolize the main arteries feeding the sacral tumor: the lateral sacral, iliolumbar, and middle sacral arteries. The bilateral internal iliac arteries were not routinely embolized. Immediately after the embolization, the patient was taken to the operation room and upon the administration of general anesthetic, a balloon was placed inside the abdominal artery via femoral access in the supine position [4]. The inverted “Y” incision comprised a posterior longitudinal midline incision in the lumbosacral region with the distal end above the sacrococcygeal joint and two other incisions starting from above the sacrococcygeal joint and running along the fibers of the gluteus maximus muscle to expose the posterior aspect of the sacrum and the adjacent ilium (Fig. 1). The presacral space was exposed by removing the sacrotuberous ligament and coccyx. The rectum was bluntly dissected from the tumor up to the S1 level and gauze was packed into the space between the rectum and sacrum to position the rectum forward. From the posterior side, the vertebral lamina was removed, the sacral canal was revealed, and the sacral nerves were carefully separated. The neural foramina were enlarged near the tumor. Part of sacrum below S3 was removed to better reveal the tumor. Next, an incision was made on the posterior side of the pseudocapsule around the tumor. The tumor was dissected carefully from the interior surface of the pseudocapsule. Part of the whole tumor was first excised and then a double-strand of sutures (No.1 MERSILK, ETHICON) was applied to remove the remaining part of the tumor. By pulling the sutures fixed in the tumor, the remaining part of the tumor could be lifted up to facilitate excision of the whole tumor. This "fishing" method could be repeated until the whole tumor within the pseudocapsule was removed in a piecemeal manner. In this study, a total of 57 patients received reconstruction, that is, lumbo-iliac fixation. The lumbo-iliac fixation was performed to restore axial and pelvic stability, and achieved by inserting bilateral pedicle screws at L4 and L5 levels as well as bilateral pedicle screws (two on each side) into each ilium. The upper iliac screw was connected to the L4 pedicle screw on the same side by a longitudinal rod, and so was the lower iliac screw to the L5 pedicle screw. The indication for lumbo-iliac fixation included sacral neurogenic tumors where the upper border was above the lower rim of the sacroiliac joint and thus more than 50% of the sacroiliac joint was resected during the surgery (Fig. 2).

2.3. Postoperative care

Silicon drainage tubes were inserted for wound drainage and were not removed until the daily drainage volume was < 50 mL. Systemic antibiotics were used until the drainage tube was removed. Patients were allowed to perform active exercises using the lower extremities in bed postoperatively. The patients were followed up every six months for the first two years and once a year thereafter. For each visit, X-ray, CT scan, or MRI of the sacral region were performed on patients.

3. Results

3.1. Surgery-related information

Operations lasted for a mean of 183 ± 37 min (range, 125–230 min). The mean estimated blood loss was 1098 ± 889 ml (range, 600–4100 ml). The mean follow-up time was 65 ± 39 months (range, 12–118 months). At the last follow-up, the local recurrence rate was 7.1% (11/155) (Fig. 3). For patients showing local recurrence, no further treatment was performed as no one presented symptoms. The patients were monitored by close surveillance.

Table 2
Clinical symptoms at presentation and outcomes.

Symptom	At presentation (No. of patients)	Follow-up (No. of patients)			
		Complete resolution	Remission	Persistence	Aggravation
Pain in lower limb or sacrococcygeal region	90	80	9	1	0
Neurologic deficit	14	4	2	7	1
Bladder and bowel dysfunction	31	21	7	3	0
Palpable pain less mass	13	0	0	0	0
Identification during physical examinations	7	0	0	0	0

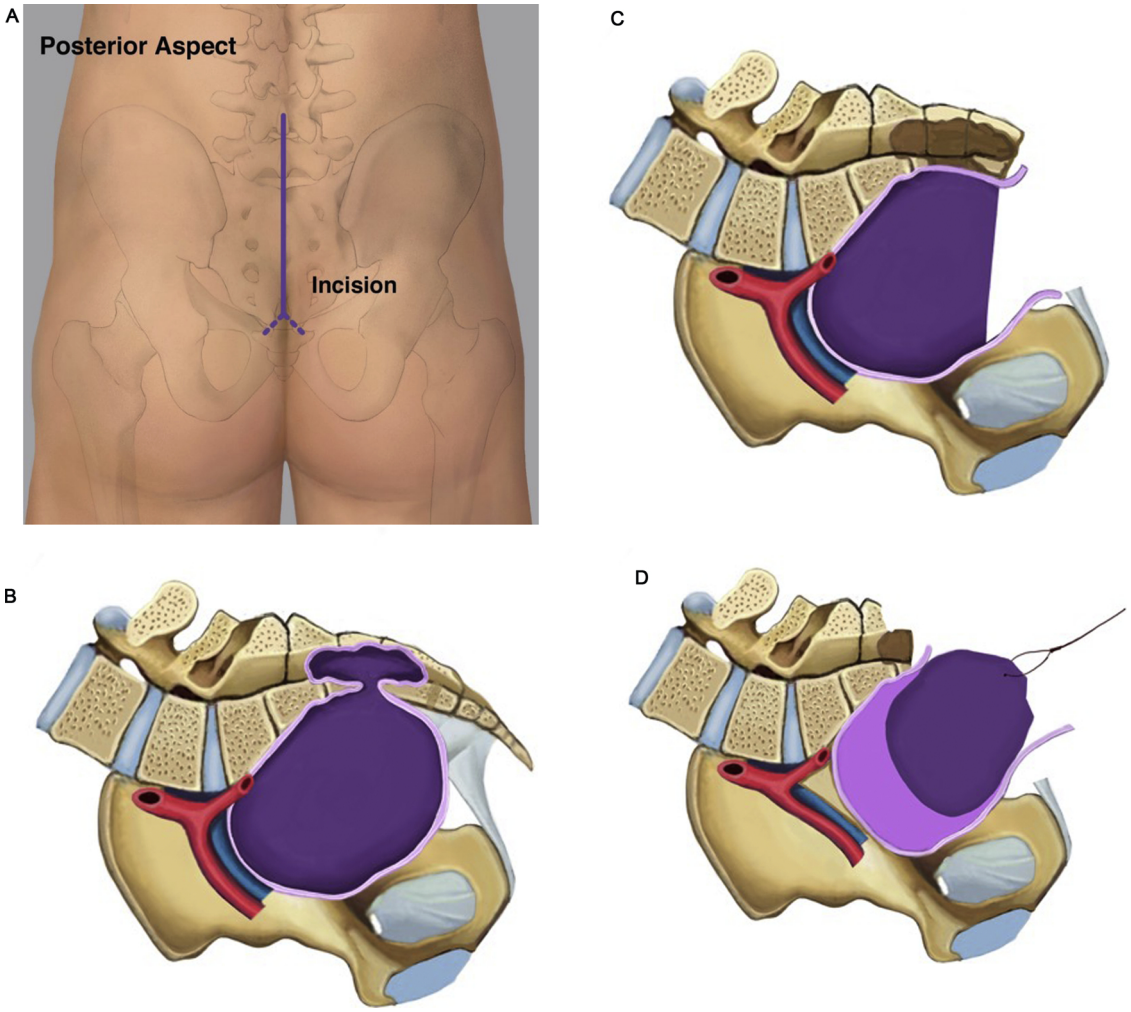

Fig. 1. Schematic diagrams showing (A) an inverted "Y" incision was used, (B) a large sacral tumor growing in the sacral canal and towards presacral space, (C) the partial sacrum below S3 and part of the whole tumor were removed, and (D) a double-strand of sutures was applied to lift up the tumor which may facilitate the excision of the whole tumor.

3.2. Symptomatic improvements

Of the 90 patients complaining of pain in a lower limb or sacrococcygeal region before the surgery, 80 experienced complete resolution of symptoms by the last follow-up, while 9 reported reduction of symptoms and one still complained persistent symptom. Therefore, 98.9% (89/90) patients experienced symptoms resolution or reduction in our study.

Through the pre-operative imaging examinations, 32 patients had tumors originating from S1 nerve roots. After the surgery, nine patients (28.1%) presented minor weakness in plantar flexion of the ankle joint, which, however, did not affect the overall lower limb function and the patients were able to ambulate without support. The other 123 patients had tumors involving S2–S4 nerve roots, of which 10 patients (8.1%) experienced various degrees of urination and defecation difficulties after the surgery, including three patients with constipation, 2 with urination problems, and 5 experiencing both bowel and urination problems. Patients who experienced constipation required more time to complete the process of defecation. On the other hand, patients who had urinary problems experienced stress incontinence, of which they had adapted by frequent voluntary urination.

Among the 31 patients who had dysfunction in urination and defecation, 21 had complete resolution, 7 had remission, while 3 presented persistent dysfunction, indicating a rate of 90.3% (28/31) for complete resolution and remission.

Of 14 patients who presented with neurological deficits before the surgery, four had complete resolution, two had remission, seven showed persistent neurological deficits and one experienced worsening of neurological functions whereby the S1 dermatomal distribution of numbness was getting worse after the surgery. Collectively, the rate of complete resolution and remission of neurological deficits was 42.9% (6/14). Taken together, the overall symptom resolution and remission rate for all symptoms was 91.1% (123/135).

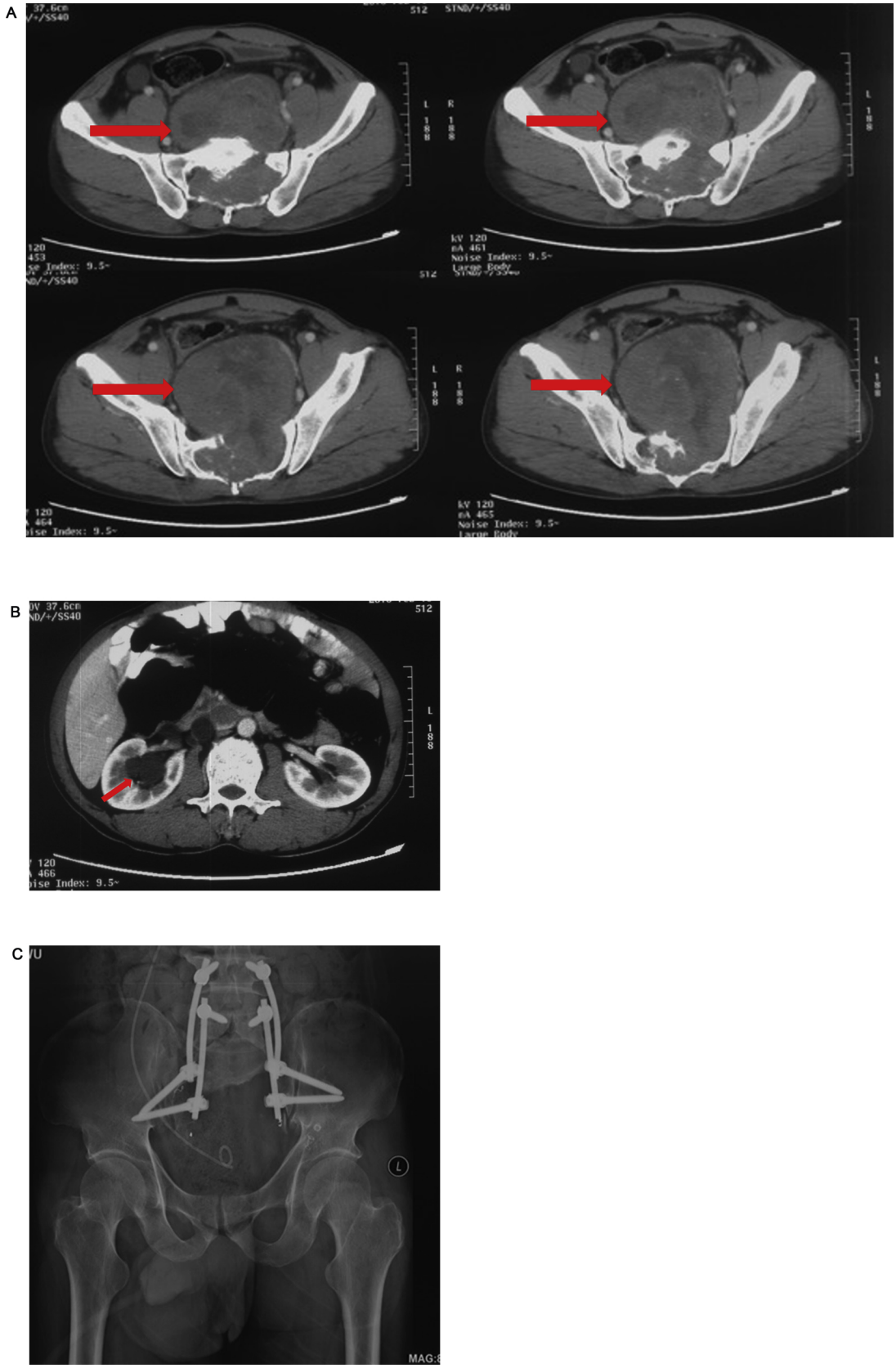

Fig. 2. (A) CT image of representative giant sacral neurogenic tumors (as indicated by the red arrows). (B) CT image of the enlarged renal pelvis (as indicated by the red arrow) resulting from the tumor compressing the ipsilateral ureter. (C) Radiograph showing reconstruction of lumbo-iliac stability after tumor resection (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article).

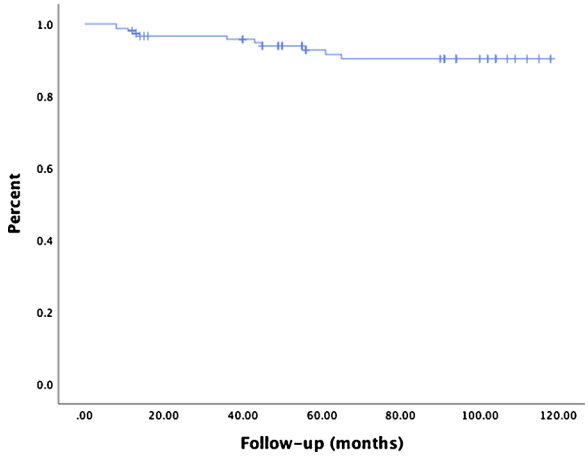

Fig. 3. Kaplan-Meier curve shows survival free of local recurrence for benign giant neurogenic tumors.

3.3. Complications

Of the 155 patients, 27 developed complications and the total rate of complication was 17.4% (27/155). Seventeen patients (11%, 17/155) experienced wound healing complications, including ten with wound healing problems and seven presenting deep infection. The ten patients that had wound healing problems were treated by debridement, and the wounds eventually healed between three and four weeks. The seven patients who developed deep infections were treated by debridement, drainage, and systemic antibiotics. The wounds eventually healed and the infections cleared. Eight (5.2%, 8/155) patients presented postoperative cerebrospinal leakage. All the patients were successfully treated with bed rest in a horizontal position without a pillow for three to five days until the drainage fluid was pink or clear, and then with the head of the bed elevated to approximately 20-30°. One patient (0.6%, 1/155) developed a distal deep vein thrombosis and resolved upon the treatment with anticoagulant drugs for six months. One (0.6%, 1/155) developed mechanical breakdown at 14 months after the surgery and was managed by the removal of the hardware.

4. Discussion

Neurofibromas and schwannomas are benign in nature, and thus treatment is not always necessary. Specifically, treatment is reserved for patients presenting symptoms. Surgical resection is the ideal treatment option to relieve the compression of tumors on the rectum, bladder, ureter, or adjacent nerves. Several reports on benign neurogenic tumors have been published in the literature [2,5–8] and are compared with our findings from this study in Table 3. Three approaches are generally used for surgical removal of benign neurogenic tumors: anterior, posterior, and combined anterior-posterior. The selection of the surgical approach is based on the location and size of the presacral tumors. Either an anterior or combined anterior-posterior approach is preferred for large tumors, and also for those with the proximal border is above S4. The benefits of these two approaches include better visualization of the pelvic structures (ureters, uterus, rectum, or bladder) and superior bleeding control. However, for large and deep-seated tumors, potential complications can occur due to inability to attain good exposure intraoperatively. For published case series whereby an anterior or combined anterior-posterior approach were used, the reported intraoperative complication including venous injuries and pelvic organ injuries, the complication rates ranged from 3.7%–58.8% [2,5,6]. On the other hand, the posterior approach is preferred for smaller tumors and for those not growing beyond S4. The advantages of the posterior-only approach include optimal visualization, the possibility of resecting tumors which locate caudally, minimal morbidity, and reductions of pelvic structure injuries. Here, by reviewing our experiences on 155 patients between 2008 and 2018, we showed that the posterior-only approach worked decently for large benign neurogenic tumors (the mean tumor size of 11.1 ± 3.6 cm (ranged 5–21 cm)) with the proximal border below S1. Specifically, our surgery procedure involved several important steps: lower partial sacrectomy (removing the sacrum below S3), performing tumor dissection via the inner surface of the pseudocapsule, and tumor dissection by placing large sutures (“fishing” technique). Using this posterior-only approach, no intraoperative complication was encountered.

The greatest risk of surgery is massive intraoperative bleeding resulting from the injuries of presacral venous plexus and arteries feeding the tumor [9]. Various methods have been applied to reduce intraoperative bleeding, such as internal iliac artery ligation, interventional vascular embolism, and usage of aortic balloon occlusion [2,10]. To control intraoperative bleeding, we used an aortic balloon to selectively embolize the main arteries feeding the sacral tumor, and dissected the tumor through the inner surface of the pseudocapsule using a posterior approach to avoid injury to the presacral venous plexus. In the present study, all the patients were stable perioperatively and the mean blood loss was 1098 ± 889 ml.

Because these tumors are benign, extensive or marginal resections are not necessary in all patients. The aim of surgery is mainly to relieve the symptoms. To date, only one study reported the postoperative status of 17 patients who underwent benign presacral tumor resection [5]. In our hospital, we performed dissection through the inner surface of the tumor pseudocapsule to maintain the continuity of the affected nerve root as much as possible. The overall symptom remission rate was 91.1%. The symptom remission rate was 98.9% for patients with lower limb or sacrococcygeal region pain, 42.9% for those with neurologic deficit, and 90.3% for those with dysfunction of urination and defecation. The local recurrence rate was 7.1% (11/155), lower than several other studies [2,5,6]. No further treatment was performed on patients presenting local recurrence as there were no symptoms. Nine patients (9/32, 28.1%) with tumors originating from the S1 nerve roots developed weak plantar flexion in the corresponding ankle joint, and 10 out of 123 (8.1%) patients with tumors from the S2-S4 nerve roots experienced different degrees of urination and/or bowel difficulty.

The reported rate of postoperative complications after presacral neurogenic tumor resection ranged between 17.6% to 43.8% [2,5–8]. Many factors could contribute to this rate, including a prolonged operating time, large surgical exposure, wound in proximity to the anus, the presence of a huge dead space after tumor resection, nerve injuries, and poor blood supply to the skin flap [11–14]. The complication rate in our study was 17.4%, including 11% of patients developing wound healing problems. The relatively low complication rate may result from short surgical time (mean 183 ± 37 min) and the fact that we did not ligate the internal iliac arteries that may help to preserve blood supply to the skin flap.

In conclusion, although certain functional loss resulting from surgery is inevitable due to the location and characteristics of giant benign sacral neurogenic tumors, patients with symptoms could still benefit from tumor resection using a posterior-only approach.

Table 3
Comparison between the current and previous studies on presacral neurogenic tumors.

Study	Hébert-Blouin et al. [5]	Sun et al. [6]	Chen et al. [7]	Guo et al. [2]	Lee et al. [8]	Current study
No of patients	17	64	16	48	1	155
Mean follow-up time (months)	36	58.2	59	47	24	65
Mean operation time (min)	NR	180	252	180	NR	183
Surgical approach	anterior (N = 13); combined anterior-posterior (N = 4)	anterior (N = 19); posterior (N = 25); combined anterior-posterior (N = 20)	Posterior (N = 16)	anterior (N = 7); posterior (N = 22); combined anterior-posterior (N = 19)	posterior	posterior
Mean tumor size (cm)	7.4	NR	17.5	12	8.6	11.1
Tumor type	benign	benign	benign (N = 12); malignant (N = 4)	benign (N = 41); malignant (N = 7)	benign	benign
Blood loss	1043 ml	878 ml	1293 ml	1600 ml	NR	1098 ml
Method used to reduce bleeding	none	intra-arterial embolization	intra-arterial embolization	BDC or ligation of unilateral internal iliac artery	none	intra-arterial embolization and BDC
Local recurrence rate	11.80%	12.50%	6.30%	10.40%	0	7.10%
Rate of symptom improvement or resolution postoperatively	84.6% (11 in 13 patients with symptoms)	NR	NR	NR	0	91.1% (123 in 135 patients with symptoms)
Intraoperative complication rate	venous injuries 58.8%	6.3% (Injuries of the internal iliac vein and its branches 4.7%; ureteral injury 1.6%)	NR	3.7% (hemorrhagic shock and DIC 1.6%; rectum injury 2.1%)	0	0
Postoperative complication rate	17.6% (hemorrhage 5.9%; ileus + acute respiratory distress syndrome + deep vein thrombosis 5.9%; transient foot drop 5.9%)	35.9% (wound healing problem 15.6%; postoperative cerebrospinal fluid leak 4.7%; bowel and bladder dysfunction 12.5%; lower limb motion dysfunction 3.1%)	43.8% (wound healing problem 25%; bowel and bladder dysfunction 18.8%)	39.2% (wound healing problem 14.9%; postoperative cerebrospinal fluid leak 10.6%; bowel and bladder dysfunction 6.4%; thrombosis caused by the BDC 3.1%; retroperitoneal hematoma 2.1%; vesicorectal fistula 2.1%)	0	17.4% (wound healing promblem 11%; postoperative cerebrospinal fluid leak 5.2%; deep vein thrombosis 0.6%; mechanical breakdown 0.6%)

NR: not recorded; BDC: balloon dilation catheter.

Declaration of Competing Interest

On behalf of all authors, the corresponding author states that there is no conflict of interest.

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