Surgical Management of Giant Sacral Schwannoma: A Case Series and Literature Review

Kyoichi Handa[1], Hiroshi Ozawa[2], Toshimi Aizawa[1], Ko Hashimoto[1], Haruo Kanno[1], Satoshi Tateda[2], Eiji Itoi[1]

OBJECTIVE: **Giant sacral schwannomas are rare and difficult to treat. Unexpected neurologic deficits after surgery and tumor recurrence should be considered when surgery is performed. We attempt to remove the tumors via piecemeal total or subtotal excision, leaving parts of the capsule adjacent to nerves to preserve the nerves. This study aimed to present the cases of giant sacral schwannoma at our institutions as well as review the relevant literature and to discuss surgical management.**

METHODS: **This study included 11 patients (5 male and 6 female, mean age 53 years) with giant sacral schwannoma who were treated surgically. The clinical features, surgical details, and outcomes were investigated retrospectively.**

RESULTS: **The tumors were intraosseous type in 4 cases, dumb-bell type in 3 cases, and retroperitoneal type in 4 cases. The surgeries were performed by a combined anterior and posterior approach in 4 cases, a posterior approach in 4 cases, and an anterior approach in 3 cases. The tumors were removed via piecemeal total or subtotal excision in 7 cases, partial excision in 3 cases, and enucleation in 1 case. After surgery, symptoms improved in all cases. The tumor recurred in 2 patients after partial excision. No patients receiving piecemeal total or subtotal excision showed recurrence. One patient developed motor weakness after piecemeal subtotal excision, but the symptoms resolved.**

CONCLUSIONS: **Adopting an appropriate surgical approach based on the location of the tumor is important. Piecemeal total or subtotal excision, with parts of the capsule adjacent to nerves left behind may help achieve a good outcome, avoiding a postoperative neurologic deficit.**

INTRODUCTION

Schwannomas are benign, encapsulated neoplasms arising from the myelinated nerve sheath. They commonly occur in the thoracic region, followed by the cervical and lumbar regions.[1] Schwannomas in the sacral regions are rare,[2-4] accounting for 1%–5% of all spinal schwannomas.[4] The diagnosis of sacral schwannomas often is delayed due to its rare incidence and nonspecific symptoms. As a result, we occasionally encounter giant tumors.

Although giant sacral schwannomas are benign tumors, they are locally invasive to the surrounding organs. If neurovascular structures are located near the tumor, surgical treatment can be difficult. Indeed, some patients have demonstrated neurologic deficits after total excision.[5,6] However, partial excision has resulted in recurrence in other cases.[2,6] A consensus regarding whether tumors should be excised totally or partially has therefore yet to be reached.

We recently attempted to remove giant sacral schwannomas via piecemeal total or subtotal excision, leaving parts of the capsule adjacent to the nerves for the preservation of the nerves. The purpose of this study was to present the cases of giant sacral schwannoma at our institutions as well as review the relevant literature and to discuss their surgical management.

MATERIALS AND METHODS

This study included 11 patients with giant sacral schwannoma who were treated surgically at our institutions between 1997 and 2016. There were 5 male and 6 female patients, and the mean age was 53 years (range 35–73 years). The average follow-up period after surgery was 50 months (range 12–84 months). Follow-up was completed in 8 cases because no evidence of recurrence was observed for 60 months after surgery; 3 cases are still undergoing follow-up. The tumors were diagnosed by a pathologic examination. Giant sacral schwannoma was defined as a longitudinal length of the tumor ≥50 mm on computed tomography or

Key words
- Giant schwannoma
- Pelvis
- Sacrum
- Schwannoma

Abbreviations and Acronyms
MRI: Magnetic resonance imaging

From the [1]Department of Orthopaedic Surgery, Tohoku University Graduate School of Medicine, Miyagi; and [2]Department of Orthopaedic Surgery, Tohoku Medical and Pharmaceutical University, Faculty of Medicine, Miyagi, Japan

To whom correspondence should be addressed: Kyoichi Handa, M.D. [E-mail: khanda@med.tohoku.ac.jp]

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Table 1. Clinical Summary of Patients

Case	Age (years), Sex	Symptoms	Duration* (months)	Preoperative Embolization	Finding, Size (mm)	Surgery	Operative Time (minutes)	Blood Loss (g)	Complications	Outcome (follow-up in months)
1	57, F	Left leg pain	2		Intraosseous type, 55	Posterior, enucleation	174	483	None	NER (61)
2	38, M	Right gluteal pain, right leg numbness	276	A	Dumb-bell type, 110	Combined,† subtotal	529	5301	Motor weakness	NER (38)
3	44, F	Right leg pain, right calf numbness	144	AE	Dumb-bell type, 110	Combined, partial	—	—	Causalgia	Recurrence (84), second surgery, NER (78)
4	43, F	Right gluteal pain, right leg numbness	36	AE	Retroperitoneal type, 70	Combined, subtotal	633	2210	None	NER (60)
5	58, M	Right gluteal pain, right calf numbness	132		Intraosseous type, 70	Posterior, partial	553	1080	None	NER (60)
6	49, F	Right buttock and leg pain, pollakiuria	276	AE	Retroperitoneal type, 125	Anterior, total	319	3790	None	NER (60)
7	51, M	Pollakiuria	36	AE	Retroperitoneal type, 75	Anterior, total	323	2831	None	NER (71)
8	63, F	Left gluteal pain, left leg numbness	168	AE	Dumb-bell type, 70	Combined, total	434	1720	None	NER (59)
9	35, M	Dyschezia	156		Retroperitoneal type, 75	Anterior, partial	680	13700	Massive bleeding	Recurrence (30), Observation
10	73, M	Right gluteal pain, right leg pain	60		Intraosseous type, 75	Posterior, subtotal	388	2800	None	NER (12)
11	73, F	Low back pain, buttock pain	168		Intraosseous type, 50	Posterior, subtotal	310	240	None	NER (15)

F, female; NER, no evidence of recurrence; M, male; A, angiogram; AE, angiogram with embolization.
*Duration from onset of symptoms until surgery.
†Both anterior and posterior approaches.

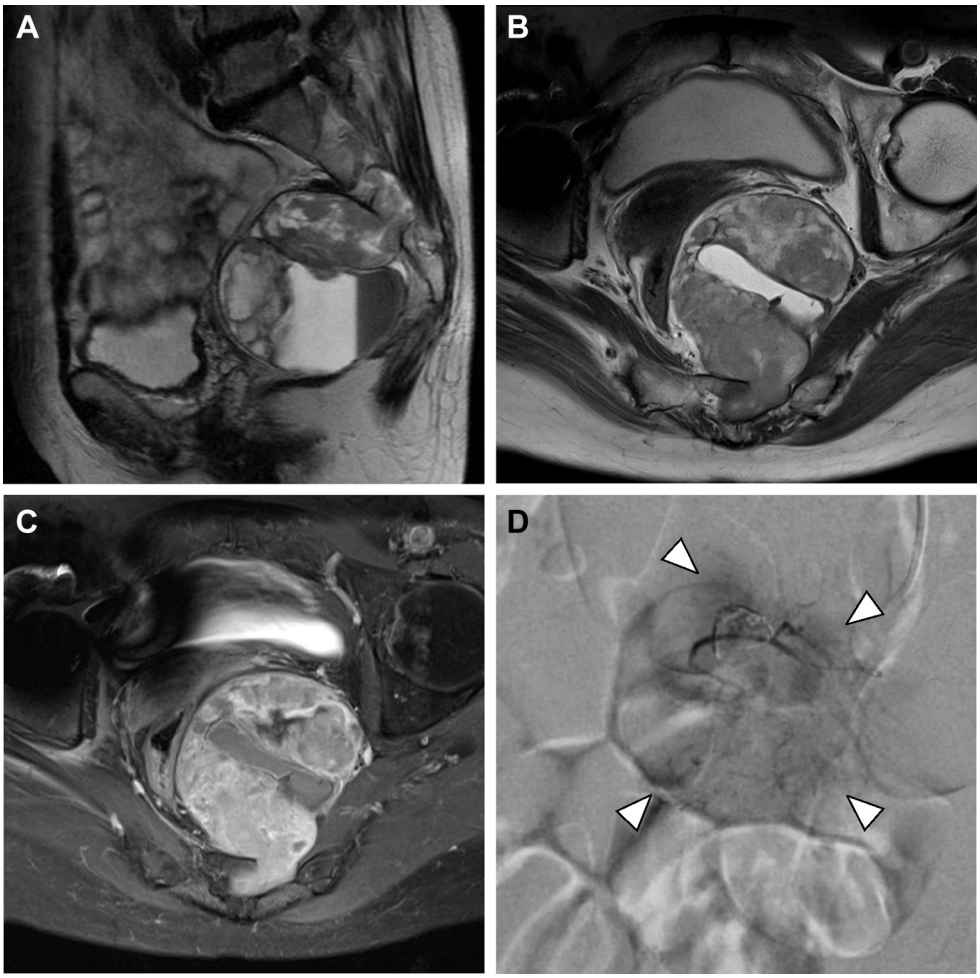

Figure 1. Case 8. (**A**–**C**) Magnetic resonance imaging showing a giant sacral tumor originating from the left S3 nerve root. (**A**) Sagittal image and (**B**) axial image on T2-weighted imaging show heterogeneous low or high intensity respectively with a cystic component. (**C**) The tumor was enhanced after gadolinium-diethylenetriamine penta-acetic acid administration. (**D**) A preoperative angiogram showing feeding arteries from the inferior gluteal artery (*arrowheads*).

magnetic resonance imaging (MRI). We reviewed the medical records and imaging studies retrospectively. Due to the data being deidentified, no ethics committee approval was needed.

Clinical Features

Symptoms, duration from the onset of symptoms until surgery, location, and size of the tumor were investigated. The location and size of the tumor were evaluated on computed tomography or MRI. The locations of the tumors were classified into intraosseous type, dumbbell type, and retroperitoneal type.[2] The intraosseous type was an intraspinal mass or an intraspinal lesion with erosion into the vertebral bodies.[7] The dumbbell type was an intraspinal mass with extraspinal extension through the sacral foramina.[1] The retroperitoneal type was located in the retroperitoneum without an intraspinal portion. The longitudinal length of the tumor was measured as the size of the tumor. If the tumor was the dumbbell type, we measured the larger part as the size of the tumor.

Surgical Details and Outcomes

The preoperative angiogram findings, tumor resection procedure, surgical approach, operative time, blood loss, surgical complications, and outcomes were investigated. The procedures for tumor resection were piecemeal total or subtotal excision, with parts of the capsule left behind to preserve the nerves, partial excision or nucleation. The part of the capsule left was determined to be the osseous side of the tumor in front of the sacrum in presacral tumors. Subtotal excision was defined as $\geq$90% of the tumor being removed but with total removal impossible. Partial excision was defined as $<$90% of the tumor being removed. The surgical approach was classified into the anterior approach, posterior approach, or the anterior and posterior approaches combined.

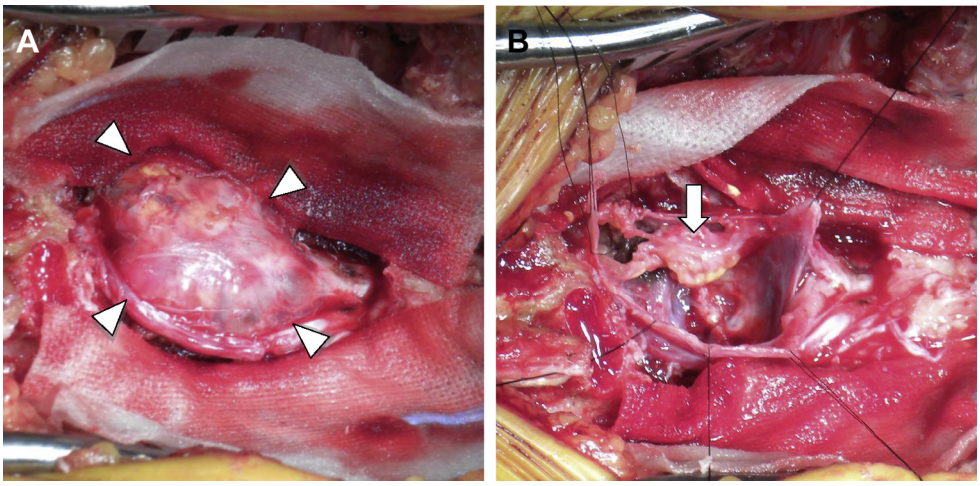

Figure 2. Case 8. (**A, B**) Intraoperative photograph during posterior approach. (**A**) The tumor originated from the left S3 root nerve (*arrowheads*). (**B**) The tumor was removed via piecemeal total excision leaving parts of the capsule of the left S3 root nerve (*arrow*).

RESULTS

The clinical features and surgical details of the patients are summarized in Table 1.

Clinical Features

Concerning symptoms, 7 patients had pain in the buttocks, 8 had leg pain or numbness, and 3 had obstruction of urination and defecation due to the bladder and rectum being compressed severely by the tumor. The duration from the onset of symptoms until surgery ranged from 2 to 276 months (average: 132 months). Regarding the location and size of the tumors, 4 cases were intraosseous type with a mean diameter of 63 mm (range: 50–75 mm), 3 cases were dumbbell type with a mean diameter of 113 mm (range: 70–150 mm), and 4 cases were retroperitoneal type with a mean diameter of 114 mm (range: 70–185 mm).

Surgical Details and Outcomes

A preoperative angiogram was performed in 6 cases, 5 of whom underwent embolization. The tumors were removed via piecemeal total or subtotal excision, with parts of the capsule left behind for preservation of the nerves in 7 cases, a partial excision in 3 cases, and enucleation in 1 case. In the intraosseous type, the posterior approach was performed in all cases. In the retroperitoneal type, the anterior approach was performed in 3 cases, and the anterior and posterior combined approach was performed 1 case. In the dumbbell type, the anterior and posterior combined approach was performed in all cases. The average operative time was 434 minutes (range: 174–680 minutes). The mean blood loss was 3889 g (range: 240–13,700 g). Regarding the surgical complication, 1 patient who had their tumor removed subtotally experienced motor weakness after surgery, but the symptom recovered after 2 years (case 2). One patient who had the tumor partially removed had causalgia that was treated by a caudal block (case 3). One patient with a retroperitoneal tumor suffered massive blood loss during surgery (case 9) because the branch of the internal iliac artery located at the bottom of the tumor was injured accidentally.

After surgery, symptoms existing before surgery improved in all cases. Two tumors treated by partial excision showed regrowth (case 3 and 9), and a second surgery was performed 7 years after the first surgery in 1 case (case 3). No case of piecemeal total or subtotal excision showed recurrence.

Case Illustrations

Case 8. A 63-year-old woman had experienced left gluteal pain and left leg numbness for 14 years. MRI showed a sacral tumor located at the sacral canal, the left S3 foramen, and the retroperitoneal space. It was of dumbbell type with its largest diameter was 70 × 65 mm (Figure 1A–C). The tumor showed heterogeneous low or high intensity on T1- and T2-weighted imaging, respectively, and was enhanced well after gadolinium-diethylenetriamine pentaacetic acid administration. A preoperative angiogram was performed, and the feeding arteries were embolized before surgery (Figure 1D). During surgery via an anterior and posterior combined approach, the tumor was removed through piecemeal total excision, leaving parts of the capsule of the left S3 root (Figure 2A–B). The patient's symptoms disappeared after surgery. There was no neurologic deficit after surgery, and no recurrence was observed during the follow-up period.

Case 9. A 35-year-old man presented with an abdominal mass and obstruction of urination. MRI showed a huge sacral tumor extending from the left S1 foramen to the retroperitoneal space and occupying a large part of the pelvic cavity (Figure 3A–B). Its largest diameter was 185 × 120 mm. During surgery via an anterior approach, the patient suffered massive bleeding from a branch of the internal iliac vein when 70% of the tumor was removed (Figure 3C–D). Vascular surgeons repaired the vein. The total blood loss was 13,700 g. We abandoned total resection. The patient's symptoms disappeared after surgery. Regrowth of the tumor was observed 30 months after surgery, but there were no

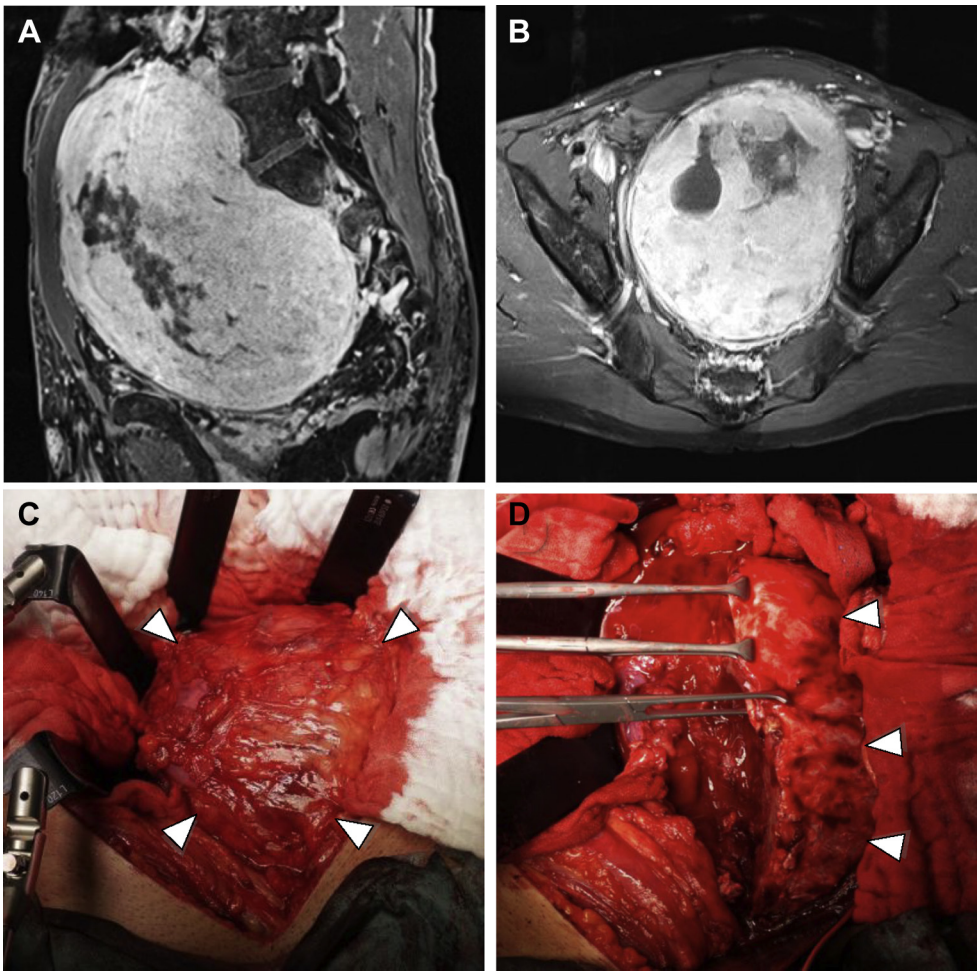

Figure 3. Case 9. (**A, B**) Preoperative (**A**) sagittal and (**B**) axial images of T1-weighted magnetic resonance imaging after gadolinium-diethylenetriamine penta-acetic acid administration. A huge sacral tumor extended from the left S1 foramen to the retroperitoneal space, and occupied a large part of the pelvic cavity. (**C, D**) Intraoperative photograph during anterior approach. (**C**) The huge tumor had many blood supplies (*arrowheads*). (**D**) The part of the tumor that remained (*arrowheads*).

symptoms (Figure 4A—B). The patient has been followed carefully.

DISCUSSION

The diagnosis of sacral schwannomas is often delayed. Abernathey et al.[6] reported that the average duration from the onset of symptoms until surgery was 5.2 years in 13 cases of sacral schwannoma. In the present study, that duration was 11 years. The reason for this delay seems to be the nonspecific symptoms of this lesion, allowing it to go unnoticed until it has grown to a substantial size, inducing compression or invading to adjacent structures. Several symptoms were observed in our cases. Seven patients had buttock pain, 8 had leg pain or numbness, and 3 had obstruction of urination and defecation. Schwannomas are slow-growing tumors, and symptoms appear once the tumor expands fully in the sacral canal or extensively destroys the sacrum. As a result, the tumors are huge when discovered.

Although schwannomas are benign tumors, giant sacral schwannomas are difficult to treat because of their varied routes of extension.[8,9] Some tumors enlarge within the spinal canal, whereas others extend to the presacral space through the sacral foramina, and still others destroy the sacral body. Concerning the surgical approach, it is important to understand the location and size of the tumor and its relationship with the adjacent structures. Sacral schwannomas were classified into 3 types in the present study: intraosseous type, retroperitoneal type, and dumbbell type. Generally, the intraosseous type can be resected by the posterior approach. The anterior approach is performed in case of retroperitoneal type, as this approach facilitates control of the vessels

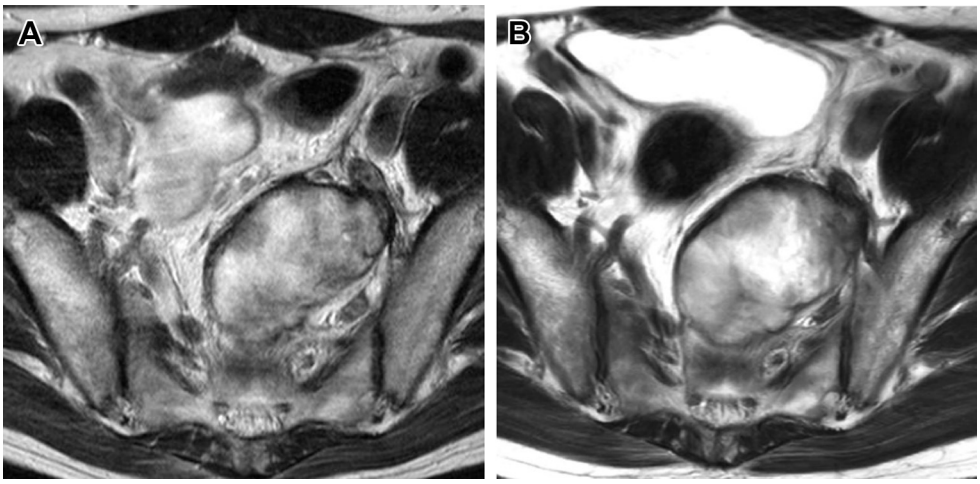

Figure 4. Case 9. Postoperative axial images of T2-weighted magnetic resonance imaging at (**A**) 6 and (**B**) 30 months after surgery. The tumor had increased in diameter from 42 × 70 mm to 47 × 75 mm.

or pelvic viscera.[5] In case of dumbbell type, the anterior and posterior combined approach typically is performed. These approaches are all reasonable for surgical treatment.

The ideal treatment is the complete removal of the tumor without neurovascular complications or spinal instability caused by bone removal.[10,11] The risk of neurologic deficits after surgery and tumor recurrence should be considered. We reviewed several articles about the surgical management, as well as the complications and outcomes of sacral schwannomas. There were 18 articles (7 case series and 11 case studies) and a total of 68 patients (**Table 2**).[2-6,8,10,12-22] Regarding complications, 7 patients (10%) had motor weakness or numbness, and 4 patients (6%) had bowel and bladder dysfunction. Regarding the outcomes, 12 patients (18%) developed recurrence, 6 of whom (9%) required reoperation. Regarding tumor resection, in some reports of total excision resulted in neurologic deficits after surgery,[5,6] and other reports of partial excision have described cases of subsequent recurrence.[2,6] Therefore, there is no consensus regarding whether these tumors should be excised totally or partially. Most schwannomas are slow-growing tumors, but some glow rapidly and occasionally transform into malignant lesions.[21,23,24] If the tumor recurs, a second surgery may be more difficult due to anatomical changes and adhesions. It is therefore best to excise as much of the tumor as possible at the first surgery. Pan et al.[22] reported that 10 patients who underwent piecemeal subtotal excision had a good prognosis after surgery. In the present study, piecemeal total or subtotal excision leaving parts of the capsule was performed in 8 cases, and none experienced recurrence. In contrast, partial excision was performed in 3 cases, and 2 experienced recurrence. Piecemeal total or subtotal excision leaving parts of the capsule adjacent to nerves may result in a good outcome, avoiding unnecessary neurologic deficit.

Some reports have described the usefulness of intraoperative neurofunctional monitoring systems, such as motor-evoked potentials, electromyography, anal pressure monitoring, and bulbocavernosus reflex monitoring.[4,25] We did not use these systems in the present case series; however, they may contribute to preventing neurologic deficits.

In surgical management, preoperative embolization of the tumor-feeding arteries is recommended to decrease intraoperative blood loss.[26] However, in some cases, embolization cannot be achieved because the tumor receives a substantial volume of blood from the bilateral internal iliac arteries. The embolization of the bilateral internal iliac arteries occasionally has severe complications related to ischemia of the embolized tissues.[27,28] Indeed, in the present study, embolization failed in 1 of the 6 patients who underwent a preoperative angiogram, due to the tumor being supplied with blood from the bilateral internal iliac arteries.

We could not analyze proper surgical managements statistically due to the lower numbers of patients in the present study. However, because schwannomas in the sacral regions are very rare, it should be meaningful to inform our experience.

CONCLUSIONS

Our experience suggests that piecemeal total or subtotal excision, with parts of the capsule adjacent to nerves left behind, is likely to help achieve a good outcome, avoiding any postoperative neurologic deficit.

Table 2. Reported Cases of Giant Sacral Schwannomas

Study	No. Cases	Location (No. Cases)	Surgical Approach (No. Cases)	Resection (No. Cases)	Complications (No. Cases)	Outcome (No. Cases)
Abernathey et al., 1986[6]	13	None noted	Posterior (10) Anterior (3)	Total (4) Subtotal (9)	Prolonged wound drainage (1) Open drainage (1) Wound infection (1)	Recurrence (7), Reoperation (4) NER (6)
Turk et al., 1992[12]	1	Intraosseous (1)	Combined* (1)	Total (1)	Bowel and bladder dysfunction, motor weakness, and impaired erectile function (1)	NER (1)
Kawano et al., 1994[13]	1	Retroperitoneal (1)	Combined* (1)	Total (1)	Hypoesthesia (1)	NER (1)
Inoue et al., 1996[14]	1	Intraosseous (1)	Posterior (1)	Total (1)	None (1)	NER (1)
Domínguez et al., 1997[15]	6	None noted	Posterior (4) Combined* (2)	Total (2) Subtotal (4)	None (6)	Recurrence (3), Reoperation (1) NER (3)
Chandhanayingyong et al., 2008[16]	4	None noted	Posterior (4)	Total (2) Subtotal (2)	Fracture (1)	NER (4)
Ozturk et al., 2009[17]	1	Intraosseous (1)	Combined* (1)	Total (1)	Motor weakness and numbness (1)	NER (1)
Nishikawa et al., 2009[18]	1	Retroperitoneal (1)	Anterior (1)	Total (1)	None (1)	NER (1)
Pongsthorn et al., 2010[2]	6	Intraosseous (1) Dumb-bell (4) Retroperitoneal (1)	Posterior (2) Anterior (1) Combined* (3)	Total (1) Subtotal (3) Partial (2)	Motor weakness (1) Causalgia (1)	Recurrence (1), Reoperation (1) NER (5)
Hosaka et al., 2010[19]	1	Retroperitoneal (1)	Anterior (1)	Total (1)	None noted	NER (1)
Çağli et al., 2012[5]	13	None noted	Posterior (6) Anterior (2) Combined* (5)	Total (11) Subtotal (2)	Radiculopathy (1) Wound problem (2) Bowel and bladder dysfunction (1) Vein injury (1)	NER (13)
Yu et al., 2012[3]	5	Intraosseous (5)	Posterior (3) Combined* (2)	Total (2) Subtotal (3)	None noted	NER (5)
Kanamori et al., 2013[20]	1	Intraosseous (1)	Posterior (1)	Partial (1)	None noted	NER (1)
Togral et al., 2014[4]	1	Dumb-bell (1)	Combined* (1)	Partial (1)	Motor weakness and numbness (1)	NER (1)
Xu et al., 2015[21]	1	Retroperitoneal (1)	Anterior (1)	Total (1)	Massive bleeding (1)	Recurrence (1)
Lin et al., 2016[10]	1	Intraosseous (1)	Posterior (1)	Total (1)	None noted	NER (1)
Pan et al., 2017[22]	10	None noted	Posterior (7) Anterior (1) Combined* (2)	Total (10)	Bowel and bladder dysfunction (2) Cerebrospinal fluid leakage and secondary intracranial infection (1)	NER (10)
Khan et al., 2018[8]	1	Retroperitoneal (1)	Combined* (1)	Total (1)	None (1)	NER (1)

NER, no evidence of recurrence or regrowth.
*Both anterior and posterior approaches.

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