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Management of giant presacral schwannoma. Clinical series and literature review.

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KEY-WORDS: Presacral, Schwannoma, Giant schwannoma, Surgery, Neuroma

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ABSTRACT

PURPOSE

Presacral schwannomas are rare tumors. Due to their benign nature and slow growth, these tumors are often giant and become difficult to treat. Their removal is a surgical challenge with different strategies reported in the literature. This study presents the consecutive cases of presacral schwannomas operated on in our institution, our surgical strategy and literature review.

METHODS

This retrospective study includes all consecutive patients operated on for a pre-sacral schwannoma in our department between 2006 and 2019, i.e. 6 patients. We report clinical features, pre and post-operative imaging, surgical data and post-operative outcomes.

RESULTS

All patients had symptoms before surgery (constipation, dysuria, radicular or lower back pain) with an average duration of 7.4 months. All patients underwent an MRI and a CT scan before the surgery. Five patients had type III schwannoma according to Klimo classification and one patient had a type II. The average size was 504,9 cm³ (range 53,1-1495,4). All the patients were operated on by an anterior approach in a double team with a mean duration of 246 minutes. Intraoperative bleeding was less than 500 ml for 4 patients, 2 patients had significant bleeding (2700 and 2900ml). Excision was total or subtotal in all cases. One

patient had an intraoperative complication (air embolism). Follow up at 3 months was excellent with a disappearance of symptoms for all patients except one patient who retained constipation. One patient had a late complication (bowel obstruction due to tissue adhesions). At last follow-up after phone interview, no patient had clinical symptoms that could suggest a recurrence.

CONCLUSION

The anterior approach with a double surgical team is a great option for the treatment of presacral schwannoma. Combined with adequate preoperative imaging and intraoperative stimulation, it reduces the risk of intra and postoperative complications.

KEY-WORDS: Presacral, Schwannoma, Giant schwannoma, Surgery, Neuroma.

1. INTRODUCTION

Primary sacral and presacral lesions are very rare, benign lesions have an incidence of 0.01 cases per 100,000 people in the general population. They are mainly found in people over 70 years of age[1]. All histology combined, the presacral lesions represent one hospitalization out of 40,000[2], 34,5% are malignant tumors (metastases, teratocarcinoma, carcinoma) and 65,5% are benign tumors (cyst, fibroma, teratoma)[3,4]. One of the few causes of presacral tumors are schwannomas derived from the sacred roots. They account for 0.5 to 3% of all retroperitoneal tumors [5].

Schwannomas are benign encapsulated neoplasms that develop on cranial or spinal nerves. Spinal schwannomas are found mainly in the thoracic, then cervical and lumbar regions[6]. About 5% of spinal schwannomas develop in the sacrum [7], with different shapes according

to Klimo classification : type I (the tumor is confined to the sacrum), type II (the tumor has breached the anterior and/or posterior osseous margins of the sacrum) and type III (the tumor is located within the presacral space) [8].

Due to their benign nature and presacral location, the first symptoms of Klimo type III schwannomas usually appear when their major axis exceeds 5 cm. This size defines giant presacral schwannoma [9]. Because of their location, size and proximity to vessels and ureters, presacral schwannomas represent a surgical challenge with some controversies about the optimal approach.

The purpose of this study is to present the cases of giant presacral schwannomas in our institution and to discuss the best surgical strategy according to the literature to reduce per and postoperative complications and preserve an optimal quality of life.

2. MATERIALS AND METHODS

This retrospective study included six consecutive patients with giant presacral schwannomas who were surgically treated in our institution between 2006 and 2019. Each patient was followed up at 1 and 3 months and a final evaluation by telephone interview. Giant presacral schwannomas were defined by: a tumor diameter > 50 mm in magnetic resonance imaging (MRI), a Klimo classification of type III or II and an anatomopathological diagnosis of benign schwannoma. Data were collected retrospectively using computer and paper records. Written consent for data processing and analysis was obtained for each patient. Clinical symptoms, their duration, and a notable patient history were reported. The size and location of the tumor was determined by MRI. The volume was calculated using the equation $\pi/6 \times \text{Length} \times \text{Width} \times \text{Thickness}$. Each patient underwent a urological CT scan before surgery to

check the relationship between the tumor and the ureter. One patient (case 5) had preoperative embolization of the tumor due to its large size and significant vascularization. The surgical approach was an anterior transperitoneal approach performed in a double team, neurosurgeon and general surgeon, with the same team for all the patients. All of the tumors were removed by debulking the tumor as it was impossible to get it out as a single piece (too dangerous for the dissection of iliac vessels). Small remnants of the capsule were left on the carrier nerve to preserve it as much as possible. Dissection on the nerve was helped with electrophysiological monitoring. No microsurgical technique was needed. The ureters were visualized by their color (whitish) and their peristalsis. They were carefully isolated as soon as located.

Intraoperative electrostimulation was used in all cases to identify and protect S1 root. We performed an electromyographic recording (sural muscle) with stimulation-detection of S1. Surgical data surgical approach, operative time, blood loss, intraoperative complications, quality of resection. We report post-operative symptoms and 1 and 3 months after surgery on the digestive, urinary, sexual and neurological aspects. A postoperative MRI was systematically performed at 3 months to assess the quality of resection. We called the general practitioners and/or the patient to obtain the latest clinical outcome.

3. RESULTS

Initial clinical features, surgical management and clinical outcomes are presented in table 1.

3.1 Clinical presentation

A total of three men and three women with an average age of 52 years (range 32 - 69) underwent surgical treatment for presacral schwannoma in our department between 2006

and 2019. The mean duration of symptoms prior to diagnosis was 7,5 months (range 2 - 24). Two patients had urinary problems (dysuria). Three had radicular pain, one patient had bilateral radicular pain and two patients had unilateral radicular pain (one in the right S1 territory and the other in the left L5 territory). None of the patients had any motor or sensory deficit. The patients with radicular pain had dysesthesias. One patient had constipation. One patient had low back pain. None of the patients were known to have neurofibromatosis type 2.

3.2 Radiological presentation

A preoperative MRI was performed for all cases. Five tumors were only presacral (Klimo type III) and one infiltrated the sacrum (Klimo type II). MRI revealed a heterogeneous hyperintense T2, hypointense T1, enhanced-contrast presacral mass. All tumors were well circumscribed without peripheral oedema, in favor of their benign nature. Three schwannomas derived from the root S1, 2 from S2 and 1 from S3. Two schwannomas had a long axis greater than 15 cm, the others were between 5 and 10 cm major axis. The average volume was 504,9 cm³ (range 53,1 - 1495,4). The CT scan with contrast (early and late time) performed before surgery identified the relationship between the schwannoma, the vessels and the urinary tract. Regarding the patient with preoperative embolization, injection of the left upper hypogastric artery revealed that the tumor was vascularized by the upper and lower lateral sacral arteries. These arteries were embolized with good angiographic results.

3.3 Surgery and intraoperative complications

Resection was total in one case (case 6) and subtotal in 5 cases.

The average duration of the surgery was 246 minutes (range 90 - 580). Intraoperative blood loss was less than 500ml for 4 cases. Bleeding was significant for two cases. For case 5, there was an internal iliac vein injury, explaining 2800ml of blood loss. For case 1 (blood loss of 2900mL) it was a diffuse bleeding without any vascular injury. These two cases required a blood transfusion. Case 5 was complicated by an air embolism at the end of the intervention. The duration of the surgery and blood loss were proportional to the size of the tumor. The carrier nerve root has been preserved for all cases. The pathological examination concluded with a schwannoma in each case.

3.4 Outcomes

Patient 5 remained in intensive care for 48 hours after surgery because of the air embolism. For the other cases, there were no postoperative complications. The average length of stay was 6,16 days (range 5 - 10). Postoperative pain was controlled with paracetamol and tramadol. One month after surgery, 2 patients had no symptoms, 2 patients had isolated constipation and 2 others had hypoesthesia in the territory of the operated root. Three months after surgery all patients were asymptomatic except for case 6 which still reported constipation. The postoperative MRI performed at 3 months showed complete resection with residual contrast enhancement on the carrier root for cases 2 to 5. MRI showed complete resection with no residual contrast for cases 1 and 6. Because of the very low risk of recurrence, the patients were followed up by the general practitioner afterwards. Monitoring was clinical for all patients. After phone interview, none of them had any symptoms related to the surgery. The average length of follow-up was 67 months (range 7 - 165). One patient was operated on 132 months later for bowel obstruction due to tissue

adhesions (case 5), exploration during the operation did not reveal any recurrence of schwannoma.

4. CASE ILLUSTRATIONS

4.1 Case 1

A 67-year-old man had dysuria and constipation for about 2 years. The diagnosis was difficult, a colonoscopy was first performed due to the presence of blood in the feces. There was no radicular or lower back pain. A CT scan was finally requested by the general practitioner and the diagnosis of presacral schwannoma was suggested. The MRI revealed a large tumor in T2 heterogeneous signal with a large intra-tumor cyst, the tumor was in T1 hyposignal and was enhanced after gadolinium administration (figure 1). The size is evaluated at 17cm in height, 14cm in width and 12cm in thickness, the lesion was developed on the right S1 root. There was compression on the left sigmoid colon, which explain constipation. An angiographic and urographic CT scan was performed before surgery. The lesion compressed laterally the internal iliac arteries. The pelvic ureters which were not opacified at the late stage, indicating compression and explaining the dysuria. There were intralesional calcifications (figure 2). The patient was operated on via an anterior transperitoneal approach. The lesion was very hemorrhagic. The vessels and ureters were identified and protected in order to avoid any injury. The lesion was then opened and emptied to facilitate dissection. The entire lesion was removed except for a small portion in close contact with the left S1 root. The operation lasted about 4 hours. The amount of blood lost was 2.9L due to the hemorrhagic nature of the lesion during removal. The patient received a blood transfusion at the end of the intervention. He stayed 6 days in digestive surgery department without post-operative complications. One month after surgery the

dysuria and constipation disappeared, the patient felt tired. Evaluation at 3 months was excellent with a control MRI documenting a complete excision (figure 1) and an asymptomatic patient who had returned to normal life.

4.2 Case 2

A 34-year-old woman had right S1 root pain for 2 months for which a lumbosacral MRI was performed. A 5 x5 cm lesion developed on the right S1 was discovered. The lesion was in isosignal T2, hyposignal T1 and strongly enhanced after gadolinium administration. The diagnosis of schwannoma was considered (figure 3). An angiographic and urographic CT scan was performed before surgery. It detected close contact between the upper part of schwannoma and the superior gluteal artery. The internal iliac vein was in contact with the anterior part of the schwannoma (figure 4). Intraoperative electrostimulation was used to monitor the right S1 root. The operation was uneventful (150 minutes) and simple. Intraoperative bleeding was not significant. The patient spent 5 days in hospital. Evaluation at 1 and 3 months was excellent with a complete resolution of the symptomatology and a complete resection confirmed by MRI except a very little part on the right S1 root. The patient was operated on 6 months later for a schwannoma in the left thigh which was aesthetically disturbing. The investigation for neurofibromatosis type 2 was negative.

5. DISCUSSION

5.1 Diagnosis and imaging

Presacral schwannomas are rare tumors, often diagnosed at a late stage due to their slow growth and their benign nature. Other diagnoses are often mentioned before surgery such as gynecological lesions [10,11] or chordomas even though the radiologic features are quite

different [3]. Two cases in our series were biopsied before referring to our department to rule out the diagnosis of chordoma. The most common symptoms of presacral schwannomas are radicular pain (28.7%), followed by lower back pain (21%) and digestive symptoms (15.7% constipation and 12% abdominal pain)[9]. Sometimes these tumors are associated with local compression and induce hydronephrosis, aortic compression[12] or vein thrombosis[13]. MRI shows a T1 hyposignal and a T2 hypersignal. There are cystic, necrotic or hemorrhagic forms. The contrast enhancement of the tumor is heterogeneous and depends on the partitions and cystic parts of the lesion[14]. The preoperative angiographic and urological CT scan allows analysis of the relationship of the tumor with the sacrum, pelvic vessels and ureters (after injection at an early and late stage), in order to plan carefully surgery. CT scan also facilitates diagnosis by detecting calcifications, which can be present in most neurogenic tumors[15]. The realization of a PET-scanner with 18-FDG, although not very useful for the management, finds out an intense hyperfixation[14]. Only one of our cases underwent arteriography with preoperative embolization (case 5). The preoperative embolization issue is controversial. We consider that it has not been effective in our case, as blood loss was one of the highest in our series. However, some teams continue to use embolization before surgery and have good results [16].

5.2 Surgery

According to the literature, the majority of types III according to Klimo are operated with anterior approach, while types II are frequently operated on by a combined anteroposterior or posterior approach alone. The average complication rate by the anterior route is 36% (intra and postoperative complications) [9]. The anterior approach has the advantage of not causing spinal instability [17]. Our surgical team had chosen to operate by transperitoneal

route for several reasons. First of all, for giant schwannomas it was not possible to lift the peritoneal sac as the tumor mass is so large. In addition, this mass had to be broken up to safely dissect the iliac vessels and ureters. Moreover, it was a team practice, especially for the digestive surgeon, who had a perfect knowledge about this type of surgical approach while being an expert in laparoscopic surgery. In our series, the rate of intraoperative complications is 16% (case 5) with an air embolism occurring during the wound on the internal iliac vein. It was the first surgery with this surgical team, showing that experience is necessary to operate on this type of tumor. In the double team, the digestive surgeon performs the first operating phase with the surgical approach and the identification of vessels and ureters, the neurosurgeon, by virtue of his knowledge of the lumbosacral plexus, helps with the identification of the sacral roots and the excision of the lesion. This allows the association of two complementary specialists and seems to reduce both operating time and surgical risks. Indeed, in comparison with the literature, our surgical time seems shorter for the same route, with an average of 246 minutes compared to 440 minutes in schwannomas performed by the anterior route in the Handa series [18] and 468 minutes in Pongsthorn series [19] without the size differing significantly. Blood losses are also less significant, in the 2 cases for which they are significant (2.8 and 2.9L), they are correlated with size. They are much lower than those observed in other case series for similar sizes. For example, in the Handa series, bleeding is 2831 to 13700g for presacral schwannomas with 75 to 125mm long axis operated anteriorly [18]. The bleeding stops when the tumor is completely removed. Resection is faster and more efficient with two trained operators, effectively reducing bleeding. Malignant schwannoma are associated with severe bleeding [20].

The anterior laparoscopic approach can be an interesting alternative, especially for post-operative follow-ups, since it seems to reduce post-operative complications. However, in the

article of Kang et al. [21] the average length of stay is greater (11 days) and the neuromas are all less than 10cm from the long axis. The posterior approach is a possible alternative when it is a type II neuroma according to Klimo's classification. According to some authors it would allow better control of the vessels and a reduction in bleeding [22]. However, this strategy is not always applicable to very large schwannomas and still carries out the risk of spinal instability. The future is probably in robotic surgeries with the Da Vinci® robot which allows better visualization, using two instruments at the same time and intraoperative stimulation. The post-operative suites also seem simpler with a shorter stay and a quick return to work [23]. The intraoperative stimulation allows close monitoring of the S1 root in cases where it is in close contact with the neuroma, it is not really reported in the literature, it should, because of its simplicity of use, be used as much as possible.

5.3 Histology

The histological results all revealed benign schwannomas, but there are rare malignant forms[20]. Macroscopically, schwannomas are clear, solid, readily hemorrhagic tumors. Microscopically, there is a proliferation of spindle-shaped cells with an oval nuclei and tapered ends, with eosinophilic cytoplasm with fuzzy boundaries. The tumor cells are arranged in compact bundles (Antoni zone A) or looser bundles (Antoni zone B). Immunohistochemistry finds intense labeling for the protein S100 [24].

One of our cases (case 2) presented another schwannoma in the left sensory cutaneous femoral nerve, suggesting neurofibromatosis type 2. The excision was performed at a distance from the operation of the sacral neurinoma. Five to 18% of patients with schwannoma have another location, they are generally but not necessarily associated with

type 2 neurofibromatosis [25,26]. Our patient benefited from a complete MRI assessment, without finding any other schwannoma.

5.4 Outcomes

Our results at 3 months are excellent with no motor deficit and no sphincter disorders. The constipation in case 6 probably has a multifactorial origin. Our team therefore chose to perform a post-operative MRI at 3 months and no longer systematically monitor patients every year by delegating monitoring to the general practitioner. Surveillance was only clinical in our series which can be considered as a failure. However, recurrence is rare and is simply monitored in most cases [19,27]. One patient presented a late complication, it's a bowel obstruction due to tissue adhesion. It is a common complication of abdominal surgery, especially panproctocolectomy (15.4%), total colectomy (8.8%) and ileostomy surgery (10.6%) [28]. There are preventive measures to apply such as gentle tissue manipulation, minimizing bowel exposure and desiccation, use of foreign bodies or developing in a near future a laparoscopy/robotic approach [29].

6. CONCLUSION

Presacral schwannomas are rare, giant, late-diagnosing tumors whose removal represents a surgical challenge. The preparation of surgery with good quality imaging, the combination of several competent, experienced surgical specialties and the use of intraoperative electrostimulation allows safe and effective treatment with a low rate of complications.

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Availability of data and material: data obtained from patient's medical records

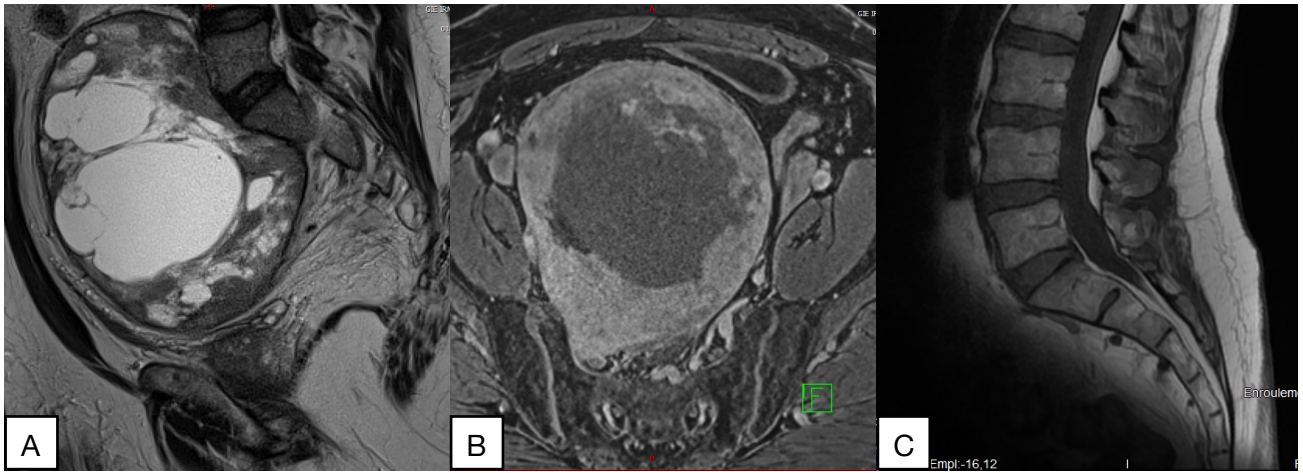


Figure 1 (Case 1) Pre-operative T2 weighted MRI demonstrate a large heterogeneous tumor with several hyperintense intra-tumor cyst (A). Post-contrast MRI T1 imaging demonstrates a peripheral enhancement of the tumor, as developed as right S1 root (B). Post operative T1 weighted MRI showing complete resection (C)

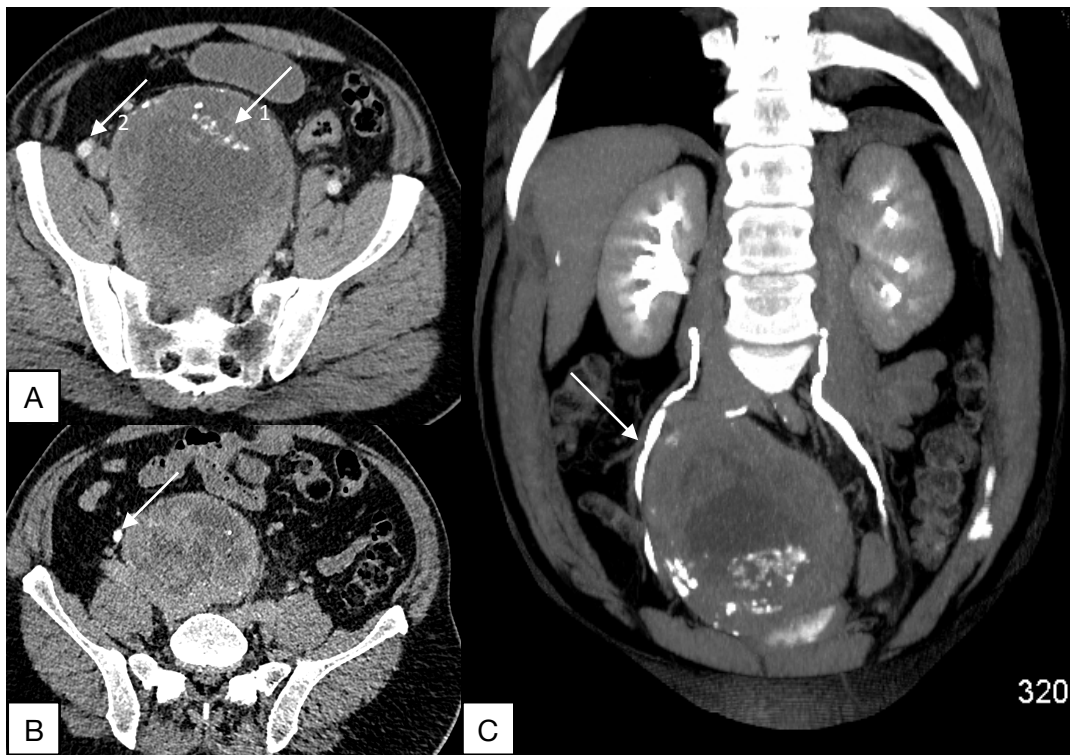


Figure 2 (Case 1) CT scan with contrast at early time (A) : arrow 1 show calcifications, arrow 2 show internal iliac artery. CT scan with contrast at late time (B) and reconstruction (C), arrows show ureter in close contact with the tumor, without dilation of the renal collecting system.

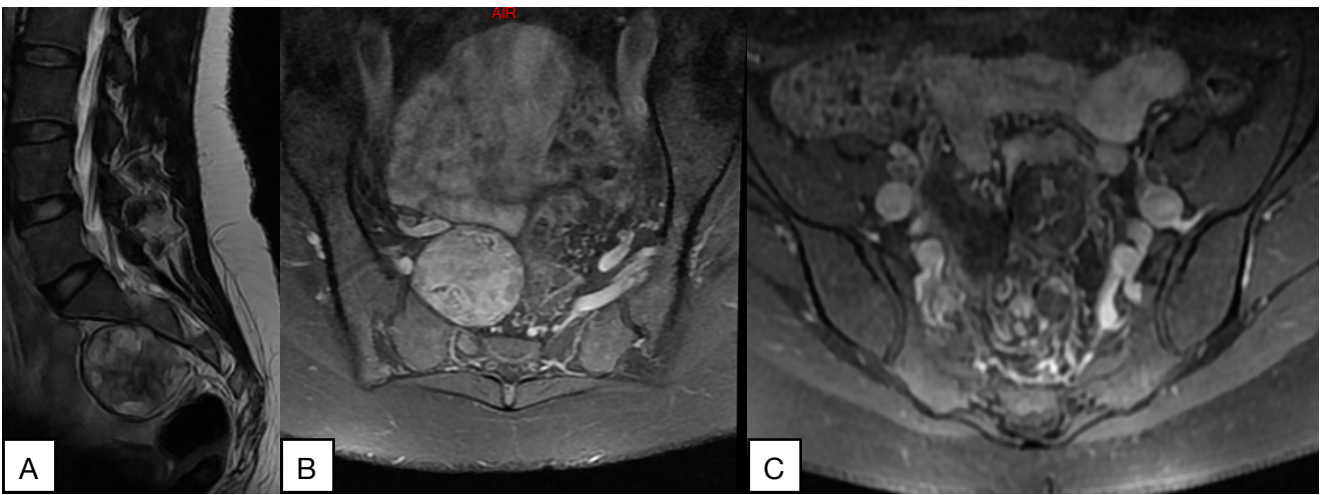


Figure 3 (Case 2) Pre-operative T2 weighted MRI (A) and T1 with gadolinium MRI (B) demonstrates a well circumscribed, enhanced presacral mass. Post operative T1 with gadolinium MRI showing no residual tumor (C).



Figure 4 (Case 2) pre operative CT scan with contrast at early time (A): arrow show gluteal superior artery, and late time (B): arrows show pelvic ureters. Pre operative CT scan reconstruction at late time (C): arrow show internal iliac vein.

Case	Age (y), Sex	Symptoms	Duration (months)	Size (length x width x thickness, cm), Location	Volume (cm ³)*	Surgical approach	Operative time (min)	Blood Loss (mL)	Resection	Intraoperative complication	Post operative complication	Length of stay (days)	Outcome 1 month	Outcome 3 months	Last outcome	Time of follow up (month)
1	65, M	Dysuria, constipation	24	17 x 14 x 12 Right S1	1495,4	Ant	240	2900	Enucleation Subtotal	No	No	6	Asthenia	Excellent	No symptom	25
2	34, F	Right S1 radiculopathy	2	5 x 5 x 4,7 Right S1	61,5	Ant	150	< 500	Enucleation Subtotal	No	No	5	Anesthesia	Excellent	No evidence of recurrence, no symptom	7
3	69, F	Lumbar pain	8	6,3 x 4,6 x 3,5 Right S2	53,1	Ant	90	< 500	Enucleation Subtotal	No	No	5	Constipation	Excellent	No symptom	8
4	32, M	Abdominal pain, Left L5 radiculopathy	3	9,2 x 7,8 x 6,3 Left S1	236,7	Ant	240	< 500	Enucleation Subtotal	No	No	6	Paresthesia of S1 territory	Excellent	No symptom	100
5	57, M	Dysuria	2	19 x 9 x 12 Left S2	1074,4	Ant	580	2800	Enucleation Subtotal	Internal iliac vein injury Air embolism	Reanimation 2 days. No hemodynamic dysfunction	10	Excellent, no symptom	Excellent	No symptom, bowel obstruction at 132 months (tissue adhesions)	165
6	55, F	Constipation, paresthesias of the lower limbs	6	7,4 x 4 x 7 Right S3	108,5	Ant	180	< 500	Enucleation Total	No	Left lower lobe atelectasia	5	Constipation	Constipation	No symptom, No evidence of recurrence	97

Table 1 : Giant presacral schwannomas operated at University Hospital of Caen, 2006–2019

Ant : Anterior, F : Female, M : Male, min : minutes, mL : milliliters, y : years, *volume given as $\pi/6 \times \text{Length} \times \text{Width} \times \text{Thickness}$