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Introduction

A schwannoma is a benign nerve sheath tumor composed of Schwann cells, than can start anywhere in the body. These tumors rarely arise within bone, among which mandible and sacrum are the most common sites of involvement. The cause of schwannomas is unknown, but they sometimes occur in individuals with certain disorders such as some types of neurofibromatosis. Because of originating in the nerve sheath, schwannomas may theoretically not induce any disturbance to the nerve’s functions, but as these tumors grow, they displaced and compress important nerve fascicles within the nerve responsible for neuropathic pain. When schwannomas develop from the sacral plexus, it may cause sciatica, vulvodynia, pudendal pain, coccygodynia, and may also affect the functions of the pelvic organs. Primary en bloc resection with tumor-free margins is therefore mandatory not at last because transformation in malignant schwannoma does exist. Management of schwannomas of the sacral nerves roots is challenging because of difficulties in accessing the lesion, risk for massive intra- and postoperative blood loss due to an extensive vascularity of the pelvis, and because of risk for surgical nerves damages. We present here a small series of three consecutive patients who underwent a laparoscopic resection of a schwannoma of the sacral nerves roots (SNR) for treatment for intractable vulvo- and coccygodynia.

Methods

Between 2008 and 2012, three women with sacral schwannoma were treated by PM. Mean age of the patients was 31 years (range 22-37 years). All patients presented with lumbosacral pain, vulvodynia nad coccygodynia from 13 months to three years, treated over years with oral and topical medicines. Because of sciatica, one patient was also treated for ankylosing spondylitis on long term by corticosteroids. All three patients had constipation and developed pollakisuria and nycturia. No had difficulty in urination. The presence of a smooth sacral masses fixed to the sacrum
was confirmed by vaginal touch. The three patients presented no neurological deficit except a reduction of sensation in the left great toe in the patient suffering from the sciatica. Neuropelvological assessment diagnosed a irritative S#2-4 sacral radiculopathy on the side of the tumor: the patients presented a unilateral positive Lasègue’s sign without signs for neurogenic nerves damages while the rectovaginal touch palpation of the lower SNR induce a trigger pain with a positive Tinel’s sign. The touch of the pudendal nerves was unremarkable in all three patients. Urodynamic testing showed bladder hypersensitivity without hyperactivity in all three patients. Transvaginal Doppler ultrasound showed in all three patients an extensive sacral osteolytic tumor mainly occupying one side of the SNR area, with mean maximal length by around 6cm with strong anatomical relationship to pelvic vessels. MRI revealed iso-density to hypointensity imaging on T1-weighted sequence and hyperintensity imaging on T2 weighted in all patients (figure 1). The tumors extended from sacral foramina #2-4 and expanded intrasacral and displaced the rectum anteriorly. The fat plain between tumor and abdominal cavity was evidenced in all cases that indicated that there was no tumor invasion into internal organs. All three laparoscopic explorations were based on the primary exposure of the sacral plexus followed by the coagulation/transection of all parametric vessels supplying and surrounding the tumor, followed by the dissection/removal of the tumor en bloc with respect of the peritumoral capsule. The procedures were performed under general anesthesia avoiding drugs that reduce muscle contractility in the administration of anesthesia. For an optimal access to the sacrum, patients were placed in a Trendelenburg position to displace the intestines out of the pelvis and into the upper abdomen. The interventions were started with the retroperitoneal dissection of the pararectal space medially to the ureter for a primary identification of the superior pole of the tumor (figure 2). The procedures were carried on with the opening of the retroperitoneal space as for a pelvic lymphadenectomy followed by the exposure of all vessels of the cardinal ligament by blunt dissection. Laterally to the internal iliac vessels, the distal portion of the sacral plexus was exposed by simple detachment of the fett-lymph-tissue. For identification or differentiation of the different sacral nerves roots (SNR), intraoperative electrical stimulation with a laparoscopic probe and a current fixed by 250 μs/35 Hz/4V was used [1]. While SNR #4 stimulation does not produce any motor reaction in the lower extremities, stimulation of SNR #3 is confirmed visually by a deepening and flattening of the buttock groove as well as a plantar flexion of the large toe and to a lesser extent of the smaller toes. Stimulation of SNR #2 produces an outward rotation of the leg and plantar flexion of the foot as well as a clamp-like squeeze of the anal sphincter from anterior/posterior. Stimulation of SNR #1 induces a motion of the leg with interne rotation while stimulation of the lumbal root #5 induces a flexion/extension of the foot. In all three patients, the tumors were located dorsally to the cardinal ligament and
present strong attachment to the SNR #2-4, the sacrum and to the coccygeal bone. All parametric vessels surrounding the tumor including the internal iliac vessels after emergence of the uterine vessels as well as the pudendal and inferior gluteal vessels were secured. Because of the age of the patients and the further desire of pregnancy, the uterine vessels were kept intact. The tumors were then detached from the SNR by traction/countertraction. Finally, the tumors were dissected from the sacral and coccygeal bone by passing through the pararectal space while all small vessels underlying the tumors were coagulated (figure 3). The tumors were liberated this way keeping the capsules intact (figure 4) and morcellated within a Lapsac to avoid contamination with any spilled tumor cells. At the end of the procedures, laparoscopic control excluded any residual bleeding and permit closure of the peritoneum. To avoid risk for postoperative bladder retention and/or bladder overdistalation, suprapubic catheters were placed inside the urinary bladder, while bladder trainings were started at second postoperative day.

**Results**

All three procedures have taken about two hours. Intraoperative blood loss were estimated less than 200ml in all patients. No any intra- or postoperative complications occurred. Bowel motion recovered spontaneously at the third postoperative. The suprapubic bladder catheters were removed in all three patients after two weeks of successfully bladder training. After a short phase of several weeks of unilateral genital numbness, all three patients recovered normal sensations. Histologic examination of the specimens showed in all three patients benign schwannomas. At the mean follow-up time of 27.66 months, no any recurrence or worsening occurred in any patients. All patients are able to walk normally without gait aids. Because MRIs control revealed no residual tumor and the patients had further desire of pregnancy, no postoperative radiations were indicated.

**Discussion**

Many studies in surgical treatment for sacral schwannoma have been reported. Most of the cases were treated by curettage and overall results were favorable due to preservation of sacral nerve roots [2-15]. Abernathey et al. reported 13 cases of schwannoma of the sacrum [7]. 54% of the patients in this study who were treated by intralesional curettage experienced tumor recurrence and underwent additional surgery. Their study suggested that schwannoma originating in the sacrum should be aggressively resected with sacral amputation and lumbopelvic fixation and that sacrifice of all or many nerve roots was required to minimize the risk of recurrence. Such
procedures might cause extensive blood loss and greater chance of having postoperative bowel and bladder dysfunction, in addition to decreased sensation and motor weakness of lower extremities due to sacral nerve roots injury. In contrast, Dominguez et al. reassured us that a conservative approach with intracapsular enucleation alone produced a favorable result of only 16 percent recurrence rate [2]. The treatment by high-dose postoperative radiation must be judged with regard to risks and benefits especially in women of reproductive age. Further follow-up studies must be conducted.

**Conclusion**

Schwannomas are very rare benign sacral tumors. In all three patients of our series, the tumors always originate from one side of the sacral foramen and extend to the adjacent structures. While lumbosacral pain with or without radiculopathy were the most frequent symptoms noted in previous studies, the predominant symptoms in our patients were the vulvodynia and the coccygodynia. The laparoscopic exploration had confirmed that no tumor has infiltrated to the peritoneum and intra-abdominal organs, as diagnosed before surgery by appearance of the fat plane. All three patients had improvement in symptoms and none of them experienced clinical worsening after surgery. Recurrence has not been evidenced in this study. Although the number of the patients and the length of follow-up are limited, we made the conclusions that laparoscopic approach of deep pelvic masses with primary control of the tumoral blood vascularity reduces considerably the risk for massive intraoperative blood loss. Primary exposure and dissection of the rectum, the ureter and the SNR make the procedure safe and easier comparing to laparotomic approach, with less risk for postoperative functional morbidities. In our own series of more than 900 laparoscopic assisted vaginal radical hysterectomies for cervical cancer, full resection of the cardinal vessels including the internal iliac arteries never induced any distal tissular ischemic damages [16]. However it is out of discussion that further clinical outcomes and sequential radiographic results are mandatory.

**References**


Figure 1: Preoperative MRI

Figure 2: Laparoscopic dissection of the left pararectal space: Exposure of the lesion

Figure 3: End situs after removal of the lesion
Figure 4: End situs: The lesion before morcellement