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Abstract

Background: Pelvic schwannomas are sporadic tumors that are not usually recognized unless they become large enough to compress surrounding organs. Since the radiological findings are nonspecific, the definitive diagnosis requires histopathological examination. The mainstay of treatment for such tumors is surgical resection, which is challenging because of their large size and retroperitoneal origin.

Case: In this report, a case of a 25-year-old male with an incidental 15 cm pelvic mass treated by a two-stage management strategy is presented. The patient first underwent endovascular embolization of the mass, and it was entirely resected without any complication after that. The postoperative period was uneventful, and the patient did not require a blood transfusion. In addition, no recurrence was detected on the control CT scan at four months postoperatively.

Conclusion: Preoperative embolization is a valuable adjunct in treating patients with hypervascular pelvic tumors, enabling a less bloody surgical resection.

Keywords: Schwannoma, pelvic mass, treatment, surgery, embolization
Introduction
Schwannomas account for approximately 5% of all benign soft-tissue neoplasms, and only 1-3% of all them are found in the pelvis (1-2). Pelvic schwannomas mainly arise from a sacral nerve or the hypogastric plexus (3-4). Most of them are benign; however, they risk malignant transformation in patients with Neurofibromatosis type 1 (5-6). Furthermore, there are no pathognomonic clinical or radiological findings for pelvic schwannomas, complicating and delaying the diagnosis and management (7). Surgical excision is the treatment of choice by which the diagnosis can be definitively established or excluded (3). Herein, we present a case of giant pelvic schwannoma that was radically resected uneventfully following endovascular embolization in light of the relevant literature.

Case Report
A healthy 25-year-old man was admitted because of a giant pelvic mass with a typical delayed presentation. The reason for his first admission to another hospital three months earlier was his high blood pressure. A renal ultrasound ordered to investigate the cause for hypertension showed an incidental giant solid pelvic mass with mild bilateral hydronephrosis. He underwent bilateral percutaneous nephrostomy to improve his impaired renal function. The MRI examination performed therein revealed a heterogeneous encapsulated 15 x 10 x 10.5 cm pelvic mass arising from the right-sided upper sacral foramina displacing and compressing the bladder, right common and external iliac artery, and vein as well as the rectosigmoid colon (Figure 1A–D). Pathological examination of the biopsy performed during the laparotomy at the second hospital was consistent with schwannoma. However, the operation had to be terminated because of the high risk for massive intraoperative bleeding and procedure-related complications of pelvic dissection. After a while, the patient was admitted to our hospital with the findings of anemia and mild renal insufficiency. On physical examination, a complex, immobile mass with a smooth surface could be palpated in the suprapubic region. There was no sign indicative of pelvic vascular compression like swelling or edema of the lower extremities. Given the large size and high mass vascularity, surgical removal following preoperative embolization was planned, informing the patient about the consequences of embolization or surgery.
Abdominal aortography was first performed via the right femoral arterial route to determine the embolization strategy, revealing a faint tumoral blush within the pelvis. Bilateral selective internal iliac arteriograms disclosed a mass mainly fed by the right internal iliac artery (IIA) without a venous shunt. After its occlusion with detachable coils, a subsequent angiogram revealed the reflux making the median sacral artery visible. Then, this was selectively catheterized and embolized using microspheres (Embosphere Microspheres; Merit Medical, Utah, USA), 300-500 microns in size, following proximal coil embolization of its branch supplying the rectum to allow collateral circulation to develop. The embolization was completed by injecting 1 mL of a mixture of monomeric n-butyl-2-cyanoacrylate glue/iodized oil (Histoacryl; Braun, Tuttingen, Germany/Lipiodol; Guerbet, Villepinte, France) at a 1:8 ratio. The final angiogram demonstrated complete tumoral devascularization (Figure 2A–D). The post-embolization course was unremarkable, except for the right-sided gluteal pain due to ischemia, which was relieved with analgesic therapy. Although the serum creatinine level elevated up to 1.78 mg/dl from a baseline level of 1.53 mg/dl, this increase was not high enough to indicate contrast-induced nephropathy. Two weeks later, the patient underwent open surgery conducted by a team including general surgeons, urologists and vascular surgeons. Preoperatively, double J stents were placed in both ureters. After having uncovered the mass occupying the whole pelvis, displacing the bladder, seminal vesicles, and rectosigmoid colon, the appendix adherent to the tumor was removed firstly. Since the rectosigmoid colon was in close contact with the mass, a low anterior resection of the rectum was conducted to allow dissection. The main feeder of the mass, the right IIA, was ligated together with the accompanying vein, following the withdrawal of the previously indwelled coils. The mass was freed of the surrounding soft tissues by a combination of blunt and sharp dissection and resected from the anterior face of the sacrum as it exited from the 2nd sacral foramen (Figure 3). An end-to-end colorectal anastomosis ended the surgery.

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The histopathological examination showed a 15 x 11.5 x 8.8 cm solid mass composed of spindle cells arranged in high and low cellular areas with a thick fibrous capsule. There were widespread areas of necrosis, hyaline degeneration, and hemorrhage. Rare atypical cells were noted. Histological staining was strongly positive for S-100 but negative for CD-34.

The patient was uneventful during the postoperative period, and blood transfusion was not required. His renal function became regular, and he was discharged seven days after the surgical operation with an analgesic prescription for pain. Four-month follow-up CT showed no abnormal findings, apart from a small fluid collection at the resection site. Although his postoperative gluteal pain disappeared with time, the patient experienced numbness in the lateral aspect of his right ankle, most probably due to nerve damage during the surgery. Erectile dysfunction, a possible complication due to the closure of IIAs, was not seen during follow-up.

Discussion
Interventional radiologists are sometimes requested to embolize tumors which cause bleeding and scheduled for resection. However, schwannomas are very rare tumors asked to be embolized. Our case is a typical example of those sufferers whose diagnosis delayed due to the insidious growing pattern of likewise tumors, which usually do not cause any complaint unless they reach a considerable size to compress adjacent structures (1-3). Indeed, most of them are noticed incidentally during imaging performed for different reasons among those between 20-40 (2). Since they may mimic various etiologies, such as fibrosarcoma, liposarcoma, ganglioneuroma, hydatid cyst, hematoma, and connective tissue diseases, making a differential diagnosis is somewhat challenging (3,6). Thus, core needle biopsy, not the fine needle aspiration, is recommended for a definitive diagnosis of schwannomas when the imaging features are not conclusive (2). Typical histological features of schwannomas include a composition of hyper- (Antoni A) and hypocellular (Antoni B) areas of spindle cells that express S-100 protein (2), as detected in this case.
Surgical excision is the method of choice for treating pelvic schwannomas (3). However, the significant risk in such operations is the damage of adjacent structures, including the pelvic viscera, ureters, relevant sacral nerves, and iliac vessels. Especially in the event of an iliac artery injury, a massive blood loss is inevitable, which may leave no choice but to cease the operation too early. Furthermore, bleeding from the sacral vessels due to the dissection of a tumor from where it attaches to the presacral fascia may lead to life-threatening bleeding. In case of massive bleeding, there are not too many alternatives, apart from packing the pelvis and giving massive transfusion. Thus, it would be more rational to take action before a hemorrhage becomes an emergency. Preoperative embolization has proved to be a valuable adjunct in treating patients with hypervascular tumors of the head, neck and spinal column (8). Furthermore, preoperative embolization can increase the ability to resect a tumor totally by downsizing and softening it, thereby reducing the procedural time. However, interestingly, the literature review showed us a number of reports presenting the use of preoperative embolization for pelvic schwannomas, most probably due to their low incidence (9-10).

In conclusion, even though it is giant and hypervascular, surgical resection of a pelvic tumor can be less bloody and challenging with the administration of preoperative embolization. In addition, it is worthy to note that initial angiographic examination exerts a great influence on procedural efficiency and safety. Another point is that one should be familiar with different types of embolic agents to ensure a safe and effective devascularization.

No conflicts of interest.

References


Figure Legends
Figure 1 [A-D]. Preoperative MRI features of the pelvic mass. A. The T1-weighted axial image shows an expansile mass filling the entire pelvis displacing the adjacent structures. The tumor is associated with the upper sacral foramina on the right side. B and C. Axial and sagittal planes of T1-weighted contrast-enhanced fat-saturated images of the pelvis revealing heterogeneous contrast enhancement within the tumor. D. The T2-weighted sagittal image demonstrates the exponential growth of the mass outward the pelvis. Note the large areas of cystic/necrotic degeneration are scattered irregularly throughout the tumor.

Figure 2 [A-D]. Digital subtraction angiography images obtained during the embolization procedure. A. The non-selective abdominal aortogram shows a faint tumoral blush within the pelvis. The right iliac arteries are displaced laterally and narrowed by the mass (arrows). B. The selective right internal iliac artery angiogram reveals heterogeneous contrast uptake within the tumor (arrows). C. The selective median sacral artery angiogram well-depicts the vasculature of the egg-shaped ostrich mass. Note the indwelling coils occluding the right internal iliac artery at its first branch (arrow). D. The final angiogram shows complete devascularization of the mass. Note the glue cast filing the proximal median sacral artery (straight arrows) and the indwelling coils within the rectal branch (curved arrow).

Figure 3. Gross pathological specimen.
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