Original Article

Surgical Outcomes of Presacral Tumor: Ten Patients' Experiences in Our Hospital

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Key Words
Presacral tumors;
Diagnosis;
Surgical strategy

Purpose. Presacral tumors are uncommon lesions with little documentation of surgical management. The purpose of this study was to evaluate outcomes and follow-up results of patients who had presacral tumors removed at our hospital.

Materials and Methods. A retrospective review of patients who had surgery for presacral tumors between January 2003 to December 2020 was carried out. Demographic characteristics, preoperative diagnostic tests, surgical procedures, histopathological results, postoperative complications, postoperative hospital stay, and outcomes were all evaluated.

Results. Ten patients with an average age of 57 ± 9.1 were studied. Bowel habit change was the most common presenting complaint (40.0 %). CT scan along was performed in 5 patients (50.0%), a combination of CT and MRI in 4 patient (40.0 %), MRI along in 1 patient (10.0 %). In 9 patients, the tumor was located below the third sacral vertebrae, but a posterior surgical approach was rarely used. The average length of follow-up for these patients was 50.6 ± 52.6 months. The average length of stay in the postoperative hospital was 6.9 ± 3.5 days. There was no postoperative 30-day mortality or unplanned 90-day readmission. Recurrence occurred in one patient, whose pathology revealed squamous cell carcinoma.

Conclusion. Tumors of the presacral space are uncommon lesions that present with nonspecific signs and symptoms, making diagnosis difficult. To optimize surgical planning, a thorough understanding of pelvic anatomy is required. Complete surgical resection is essential for patients with presacral tumors to have a longer disease-free and overall survival.

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Presacral tumors are uncommon and diverse lesions that occur in the potential space between the rectum and sacrum. Whittaker and Perberton collected 22 cases from 1922 to 1936 and discovered that the incidence was only one in every 40,000 admission patients.[1] Because of their rarity and diverse origins, these tumors are difficult to diagnose and treat. Surgeons currently use the Uhlig and Johnson classification systems.[2]

The symptoms and signs are caused by compression or invasion of surrounding pelvic viscera or nerves and they may manifest as lower back pain or perineal pain, or they may resemble various neurological defects. These tumors could also be palpated during a digital rectal examination, providing clinical clues to diagnoses. Presacral tumors, on the other hand, frequently asymptomatic; completely asymptomatic lesions occur in 26-50% of patients.[3,4] Magnetic reso-

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nance imaging (MRI) and computerized tomography (CT) provide enough information for diagnosis and treatment strategies. Because of the risk of complications such as infection and malignancy, complete surgical excision is the recommended treatment for these tumors.[5,6]

We reviewed our patients who accepted surgical treatment for presacral tumors in our hospital over an 18-year period for this study.

Materials and Methods

This retrospective study was conducted at the Department of Surgery of Chia-Yi Christian Hospital. This study looked at 10 patients who had a presacral tumor removed surgically between January 2003 and December 2020. The study excluded patients with primary or recurrent rectal cancers, as well as primary gynecologic tumors. Demographics, clinical manifestations, diagnostic methods, pathological findings, surgical procedures, and outcomes were all documented. Clinicians chose the diagnostic methods and surgical approaches. Histopathological clinicians diagnosed all patients and treated them with curative resection. Because of the risk tumor spread and other complications, preoperative biopsy was not performed routinely. Histopathology confirmed the final diagnoses. Patients were followed up on through electronic records, outpatient visits, and phone calls.

Results

10 patients (6 females and 4 males) with a mean age of 57 ± 9.1 years were studied. The patients' body mass index was 22.3 ± 2.4 kg/m^2. Bowel habit change was the most common presenting complaint (40.0%). A palpable mass was detected in 6 patients (60.0%) during a digital rectal examination. The mass was discovered by chance in two patients who were asymptomatic. Among the 10 patients with diagnostic imaging, CT scan along was performed in 5 patients (50.0%), a combination of CT and MRI in 4 patient (40.0 %), MRI along in 1 patient (10.0 %). Table 1 shows demographic information as well as clinic and radiologic evaluation details.

The average duration of symptoms was 6.4 months, with two asymptomatic patients excluded. The average tumor diameter was 6.7 ± 4.1 cm. Nine patients (90%) were located below the level of the third sacral vertebrae, and six patients (66.6%) underwent anterior approach surgery. Only one patient was treated using the combined approach, and two patients were treated using the posterior approach. As surgical treatment, three different operative approaches were used, and surgical methods were determined by clinicians. The average time for an operation was 135 ± 46 minutes (range, 60-225 minutes). The average length of stay in the postoperative hospital was 6.9 ± 3.5 days (range, 4-16 days). Two complications were recorded, one of which was surgical wound infection. The average length of follow-up for these patients was 50.6 ± 52.6 months. Table 2 summarizes the clinical data of these ten patients.

Patients with benign tumors survived the follow-up period. During the follow-up period, four patients died, all of whom were in the malignant subgroup.

Patient No. 1 complained of sacral pain for 3 months, and a pelvic MRI revealed 2.7 cm × 1.4 cm lobulated heterogeneous non-enhancing masses in the presacral area (Fig. 1A). Tailgut cyst was discovered

Table 1. Demographic characteristics

Variable	Value
Age (years) mean ± SD	57 ± 9.1
Female sex	6 (60.0)
Body mass index (kg/m^2)	22.3 ± 2.4
ASA classification	
I-II	9 (90.0)
III-IV	1 (10.0)
Clinical presentation	
Bowel habit change	4 (40.0)
Pelvic and sacral pain	3 (30.0)
Asymptomatic	2 (20.0)
Palpable masses	1 (10.0)
Radiologic evaluation	
CT	5 (50.0)
CT + MRI	4 (40.0)
MRI	1 (10.0)
Mass detected on rectal digital examination	6 (60.0)

Table 2. Summary of the 10 patients

No.	Age	Sex	Pathology	Duration of symptoms (m)	DRE detectable	Diameter (cm)	Tumor location	Operative approach	Operative time (min)	Hospital stay (day)	Complication (s)	Follow-up (m)
Benign tumors												
1	64	F	Tailgut cyst	3	No	4	Below S3	Anterior	140	7	Nil	22
2	68	F	Tailgut cyst	Asymptomatic	No	5.5	Below S3	Anterior	145	5	Nil	8
3	64	M	Schwannoma	Asymptomatic	No	3.7	Below S3	Anterior	135	5	Nil	63
4	40	F	Anorectal cyst	12	Yes	3	Below S3	Posterior	110	4	Nil	163
5	56	F	Neuroendocrine tumor	25	No	4.5	Below S3	Anterior	120	4	Nil	48
Malignant tumors												
6	49	M	Neuroendocrine carcinoma	1	Yes	10	Above S3	Anterior	60	5	Nil	120
7	55	F	Metastatic carcinoma	2	Yes	5	Below S3	Anterior	185	7	Nil	45*
8	55	M	Mucinous carcinoma	3	Yes	10	Below S3	Anterior	90	8	Wound infection	11*
9	69	F	Squamous cell carcinoma	5	Yes	15.5	Below S3	Posterior	225	16	Ileus	2*
10	51	M	Squamous cell carcinoma	0.25	Yes	8	Below S3	Combined	140	8	Nil	24*

* Death in follow-up.

through histopathological examination showed (Fig. 1B-1C).

Patient No. 3 was asymptomatic when an abdominal CT for hematuria revealed a 3.9 cm presacral mass (Fig. 2A). The histopathological examination revealed schwannoma (Fig. 2C-2D). The above two patients underwent laparoscopic excision of a presacral tumor and were discharged after one week with no postoperative complications.

Patient No. 7 complained about right sacral pain for 2 months. She had history of ovarian serous adenocarcinoma with multiple nodal metastasis status post laparotomy hysterectomy, bilateral salpingo-oophorectomy and pelvic lymph node dissection by gynecologist. Abdominal CT revealed a 4.9 cm well defined heterogeneous mass at right perirectal and presacral space that compressed lower rectum (Fig. 3). Anterior approach was used and lower anterior resection was done, and the histopathological examination revealed metastatic carcinoma. Adjuvant chemotherapy was given but poor response and she died of cancer recurrence after 45 months.

Patient No. 8 had back pain for three months, and an abdominal CT revealed a 10 cm presacral mass

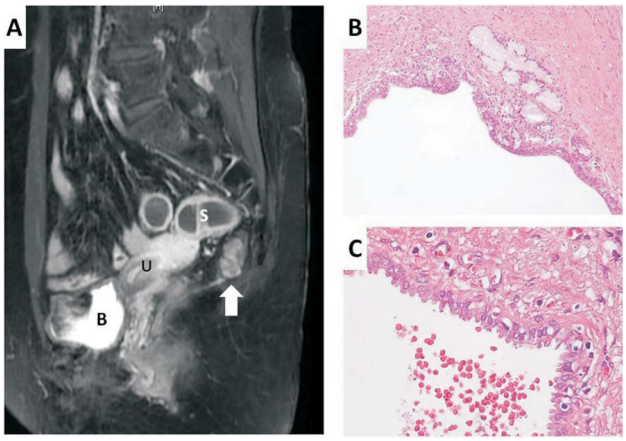

Fig. 1. Cyst of the tailgut (Patient No. 1). (A) Sagittal T2 weighted MRI of the presacral area revealed 2.7 cm × 1.4 cm lobulated heterogeneous non-enhancing masses (B: Urinary bladder; U: Uterus; S: Sigmoid). (B) Pathological examination (Hematoxylin and eosin staining at 100 × original magnification) revealing that the tailgut cyst was lined by squamous epithelium and focal columnar epithelium. (C) Hematoxylin and eosin stained ciliated squamous epithelium with gobet cells and cilia at 400 × original magnification.

with an irregular contour and coarse calcifications (Fig. 4). He also had a partial tumor resection for tis-

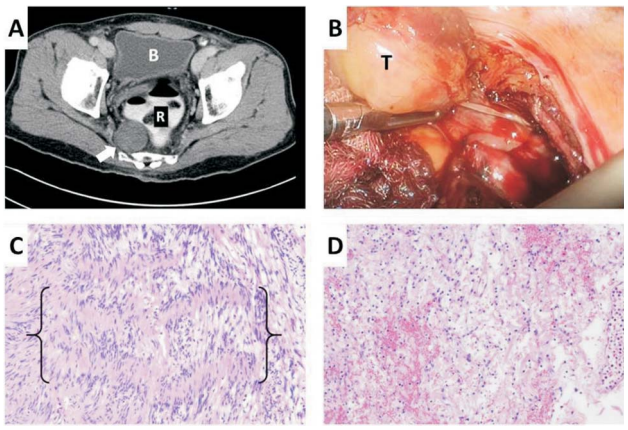

Fig. 2. Schwannoma (Patient No. 3). (A) The axial view of a contrast-enhanced computed tomography obtained during the venous phase showing a 3.9 cm × 3.5 cm homogeneous soft tissue mass (arrow) without contrast enhancement in the presacral space (B: Urinary bladder; R: Rectum). (B) The anterior approach was performed and the tumor was removed from presacral space by laparoscopy. (C) Pathological examination (Hematoxylin and eosin staining at 100 × original magnification) showing the Verocay body (bracket) with horizontal rows of palisaded nuclei separated by areas of a cellular pink basement membrane like material. Antoni A area cellular-looking area with several rows of palisaded nuclei. (D) Antoni B area with pale mucinous stroma with few cells, as well as degenerative change with focal hemorrhage, facilitating schwannoma diagnosis.

sue proof. Histopathological examination revealed mucinous carcinoma, and concurrent chemoradiotherapy was used, but the patient did not respond well. Tumor necrosis with abnormal gas formation, connecting to the lumen of the rectosigmoid colon. Unfortunately, he died after eleven months due to massive rectum bleeding. He is also the only patient who did not have a full resection.

For five months, patient No. 9 had a palpable buttock mass with tenderness (Fig. 5). For presacral mass with perineal involvement, a combined approach was used. Histopathological examination revealed squamous cell carcinoma, and she died as a result of sepsis from pneumonia.

There was no postoperative 30-day mortality or unplanned 90-day readmission. Complete resection was performed in 9 patients except the patient No. 8 as above mentioned. The disease-free survival rate, as

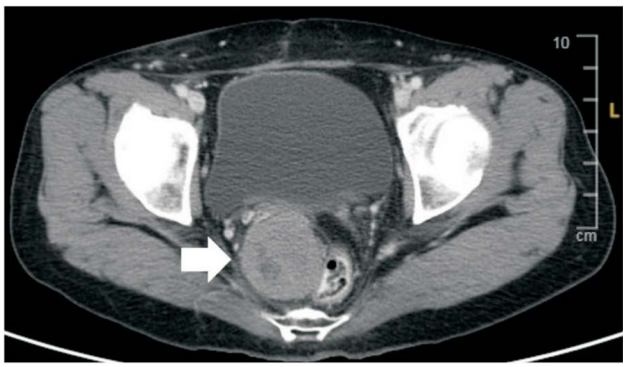

Fig. 3. Metastatic carcinoma (Patient No. 7). The axial view of a contrast-enhanced computed tomography obtained during the venous phase showing a 5.0 cm × 4.2 cm homogeneous soft tissue mass (arrow) without contrast enhancement in the presacral space.

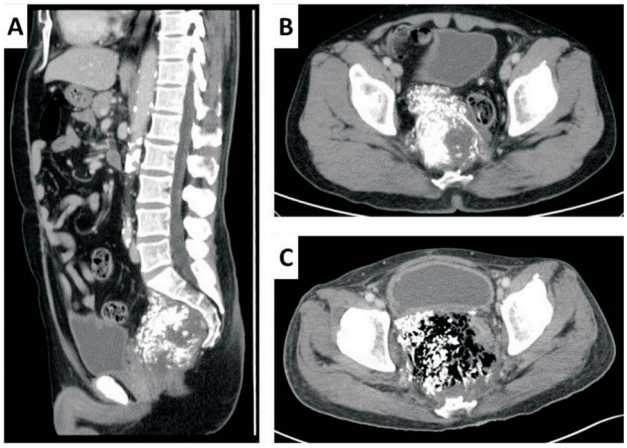

Fig. 4. Mucinous cancer (Patient No. 8). (A) Contrast-enhanced computed tomography obtained during the venous phase shows a 10 cm × 9 cm presacral mass with irregular contour and course calcifications. (B) An axial view of the tumor demonstrating the tumor adhering to the rectum in the pelvis. (C) The tumor's focal degeneration was also observed.

well as overall survival rate, at 5-year of follow-up is 55.5%. Recurrence occurred in one patient, whose pathology revealed squamous cell carcinoma.

Discussion

The retrorectal space is another name for the presacral space. It is bounded anteriorly by the rectum's fascia propria, posteriorly, by the presacral fascia overlying the sacrum, and laterally by the ureters and the

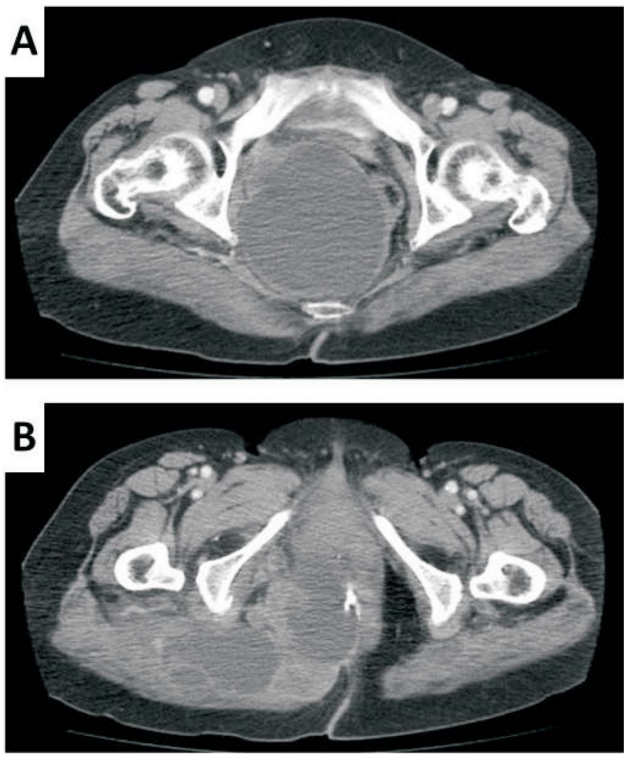

Fig. 5. Squamous cell cancer (Patient No. 9). (A) An axial view of a contrast-enhanced computed tomography obtained during the venous phase, demonstrating a 15 cm × 10 cm presacral mass that compressed to the rectum. (B) Involved muscles included the perineal and right gluteus maximus.

iliac vessels. It reaches the peritoneal reflection superiorly and Waldeyer's fascia inferiorly. Because the space is a site of development for multiple embryological structures that involute during embryonic development, it has the potential to produce a diverse range of benign and malignant tumors from the embryological remnants.

Presacral tumors are very rare with reported cases in the literature of only 1.4 to 6.3 patients would be diagnosed with this disease yearly.[2,7-9] The classification system first described by Uhlig and Johnson has been most frequently used and separates these lesions into the following broad categories: congenital, neurogenic, osseous, inflammatory, and miscellaneous.[12] These tumors are most commonly seen in female patients in their third and fifth decade of age and that the majority of these tumors can be characterized as congenital presacral tumors.[13]

Presacral tumors are typically diagnosed late due to a lack of specific clinical signs and symptoms or the fact that they are asymptomatic. The symptoms described here are caused by compression or invasion of a pelvis organ or nerve. Presacral tumor pain is dull, poorly localized, and radiating. Symptoms of presacral tumors indicate complications caused by infiltration of adjacent nerves or vessels. Pain in the lower extremities and buttocks is caused by sacral plexus infiltration.

According to Baek, digital rectal examination detected the mass in 90% of their patients.[14] Most lesions, however, are soft, compressible, and easily missed if the physician does not keep a high index of suspicion. In our study, six patients (6/10) had positive digital rectal examination results. Detailed imaging of the tumor and surrounding tissues should be an important part of the diagnosing and treating presacral tumors. CT and MRI appear to be the gold standard, with a combination of these two modalities providing the best results.[15] CT is used to show the nature of the lesion, which is cortical bone destruction. MRI is preferred over CT because it predicts benign or malignant, which is critical when planning a surgical procedure. In our study, MRI was performed in 5 patients. Hopper emphasized that MRI outforms CT (94% vs. 64%) in distinguishing benign from malignant tumor potential.[16] In our research, preoperative CT or MRI scans identified nearly all of the tumors, and the image reports were completely consistent with the surgical findings.

Historically, the role of preoperative biopsy for presacral tumors has been controversial. Traditional wisdom holds that biopsy should be avoided due to concerns about seeding along the biopsy track, and some studies have showed that needle biopsy may increase the risk of infection in presacral tumors.[7,10,11,17] Merchea discovered that preoperative fine-needle biopsy of solid presacral tumors was safe and that the results were highly concordant with those of postoperative pathology in comparison to imaging, with sensitivity, specificity, and positive and negative predictive values of 96%, 100%, 100%, and 98%, respectively.[18] Given that the goal of surgical treatment is to remove all tumors, preoperative biopsy has no utility in the management. As a result, we do not perform

preoperative biopsy in our practice.

Complete surgical resection is the cornerstone of presacral tumors management. En bloc excision allows for confirmation of the diagnosis while also removing the risk of complications such as infection, compression, recurrence, and malignant degeneration.[19,20] Imaging is used to determine the surgical procedure to be used which show the size, location, and involvement of neighboring viscera, sacrum, or pelvic sidewalls.[21] The anterior (transabdominal), combined abdominoperineal, and posterior (transsacrococcygeal or transperineal) approaches are the most commonly used for resection of presacral tumors. Tumors above the level of S3 will typically necessitate an anterior or combined approach, whereas small lesions below the level of S3 or with nerve involvement may be removed via the posterior approach, that enhanced exposure of the nerve roots provided.[22] Anterior approach provides direct visualization of the pelvic side walls and pelvic viscera, such as the iliac vessels and ureters. We believe that the anterior approach is the better method for presacral tumors, except in cases of sacrum involvement or neurogenic tumor. In our study, anterior approach was performed in 7 patients and laparoscopic surgery was applied in 2 patients despite the level of above S3. And all of the tumors were resected completely. Therefore, the S3 level should not be a contraindication to the anterior approach.

Baek reviewed previous studies demonstrating the safety of laparoscopy for treating presacral tumors. Additionally, laparoscopy offers the advantage of enhanced visualization of pelvic structures and facilitates precise dissection of the tumor from adjacent structures.[14] The three surgical approaches had no discernible difference in operative time or hospital stay day. The prognosis of patients with benign tumors receiving complete resection is excellent, the mean follow-up period was 60.8 ± 61 months.

In our patient No. 7, she had sacral pain for 3 months. She had history of ovarian cancer that received surgery resection and chemotherapy for 6 courses. During the 3 years period, there was no recurrent tumor found. The level of CA-125 was 42.6 IU/ml (normal range: 0 to 35). And metastatic carcinoma or recurrent ovarian cancer were also suspected under CT scan. For symptoms relieving and oncological outcome, laparotomy lower anterior resection was performed and postoperative adjuvant chemotherapy was given.

All patients were alive at the last time of contact. However, poor prognosis was found in patients with malignant tumors due to difficulty in performing safe margins and high recurrent rate.[7,23] In our patients, only one patient with malignant tumors was in disease free condition and the others were dead, for the 1-year survival rate was 60%. Our results support that benign tumors can be disease-free after complete resection, but malignant tumors often have a poor prognosis.

The study's limitations include a small number patients and a retrospective design. Given the diseases rarity, more research using a multicentric prospective design may be beneficial in determining the diseases natural course over time.

Conclusion

Tumors of the presacral space are uncommon lesions that present with nonspecific signs and symptoms, making diagnosis difficult. To optimize surgical planning, a thorough understanding of pelvic anatomy is required. Complete surgical resection is essential for patients with presacral tumors to have a longer disease-free and overall survival.

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原　著

薦骨前腫瘤的手術成果：單一機構的 10 例經驗

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引言　薦骨前腫瘤是一種罕見的病灶並且處理方式較少在文獻中被討論。本篇研究的目的是評估接受薦骨前腫瘤切除手術後病患的術後結果以及追蹤後續病患的情況。

方法　我們收集自 2003 年 1 月至 2020 年 12 月期間在本院接受薦骨前腫瘤切除手術患者共 10 例。回顧性地評估和分析了患者的人口統計學數據、臨床表徵、術前診斷方法、手術方式、病理組織學檢查結果、手術併發症、術後住院天數和術後結果。

結果　共 10 名患者被納入本研究，平均年齡為 57 ± 9.1 歲，最常見的症狀是排便習慣改變 (40%)。5 位患者使用電腦斷層來診斷，4 位患者合併使用電腦斷層及核磁共振檢查來診斷，只有 1 位患者單獨用了核磁共振來診斷。有 9 名患者的腫瘤位置位於薦椎第三節以下，然而會陰切除治療卻較少被使用。平均追蹤時間為 50.6 ± 52.6 個月。沒有患者在術後 30 天內死亡，並且也沒有患者在 90 天內非計畫性再入院。只有一名診斷為鱗狀細胞癌的患者發生復發。

結論　薦骨前腫瘤很罕見並且患者常出現非特異性的臨床症狀，致使診斷困難。充分了解骨盆底的結構對於優化手術計畫十分重要。完整切除腫瘤對於薦骨前腫瘤患者的無病存活期和整體存活率至關重要。

關鍵詞　薦骨前腫瘤、直腸後、診斷、手術策略。