

Original Article

# Surgical Treatment of Presacral Tumor: A Single-center Experience Over Two Decades

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**Key Words**

Presacral tumor;  
Retrorectal tumor

**Background.** Presacral tumors are relatively rare. Currently, there is no consensus regarding the appropriate surgical intervention for this disease.

**Material and Methods.** This study included patients who underwent surgery for presacral tumors at a medical center between January 2000 and December 2022. This study aimed to investigate patients with presacral tumor receiving surgery and review this rare disease.

**Result.** Data from 36 patients were retrospectively collected. Thirteen patients were male and twenty-three were female. The most common symptoms were neurological or orthopedic complaints, such as low back pain or sciatica (50%), followed by gastrointestinal complaints (27.8%). Pure laparoscopic resection was performed in 2 cases. Surgically, 14 patients (38.9%) underwent combined anterior and posterior approaches, 8 patients (22.2%) the posterior approach, and 14 patients (38.9%) the anterior approach. The estimated mean blood loss was approximately 1500 mL. The mean postoperative hospital stay was 10 days. One-fourth of the patients had a neurogenic bladder after surgery. The most common pathological diagnosis was chordoma (14 patients, 38.9%). There were more postoperative complications in tumors located above S3 vertebra than in those below S3 (66.7% vs. 11.1%,  $p = 0.004$ ).

**Conclusion.** The upper border of the tumor above the S3 vertebra is a significant risk factor for postoperative morbidity. Detailed preoperative planning and an excellent surgical strategy are necessary to ensure positive patient outcomes.

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Presacral tumors are rare heterogeneous neoplasms of diverse embryonic origins located in a potential space known as the presacral or retrorectal space.<sup>1</sup> The incidence of presacral tumors in the Taiwan population remains unknown. The incidence reported by referral centers is approximately 1 in 40,000-60,000 hospitalizations.<sup>2</sup>

The Mayo classification of presacral tumors is based on histology (benign or malignant) and origin (congenital or acquired).<sup>3</sup> Tumors in this space can have vague symptoms of various natures and intensi-

ties, depending on the tumor size and the organs involved. Symptoms usually arise when the tumor mass becomes considerably large. The chief complaints may include radicular pain, lower extremity weakness, lower back pain, incontinence, urgency, and constipation. Most commonly, the condition is asymptomatic. Thus, timely diagnosis is challenging.

Although presacral tumors are usually benign, surgical resection is generally recommended because of the risk of malignant transformation and for symptomatic relief. Since the first report by Kraske in 1885,

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much information has been published on the use of abdominal, perineal, and mixed approaches for treating such lesions. To date, there is no substantial evidence that one of these surgical methods is superior to the other.<sup>4-6</sup>

The diagnosis and treatment of presacral tumors have improved because of the development of computed tomography (CT) and magnetic resonance imaging (MRI). The choice of surgical technique depends on the size of the tumor and the tumor location in the S3 vertebra.<sup>6,7</sup> Recently, an algorithm for the surgical treatment of presacral tumors based on tumor size, bone involvement, and adjacent organ invasion has been proposed.<sup>7</sup> Neoadjuvant or adjuvant radiotherapy is used for some specific histological types but remains controversial due to the high radioresistance of tumors in this region.<sup>8</sup>

This study aimed to analyze the diagnostic approach, treatment outcomes, and postoperative morbidity of patients with presacral tumors who underwent various types of surgical interventions in a medical center.

## Material and Method

### Study population

The study enrolled patients who underwent surgical treatment for a presacral tumor at Linkou Chang Gung Memorial Hospital between January 2000 and December 2022. We retrospectively retrieved the details of each patient treated in the colorectal department from the medical records, including nursing records, operation notes, and pathologic reports.

The available medical records included age, sex, clinical symptoms, surgical strategy, intraoperative blood loss, histopathological results, and postoperative sequelae. Follow-up information was based on outpatient medical records. All patients underwent radiologic investigations, such as CT scan, MRI, or both, as preoperative evaluations to determine the appropriate surgical approach. Imaging studies were used to assess tumor characteristics, location, size, adjacent organs, bone structure, pelvic sidewall, and

vascular involvement. The extent of surgery and choice of surgical procedures were determined according to the features of the tumor.

The institutional review board of the hospital approved this study. Tumor-related clinicopathological variables included tumor location, diameter, invasion depth, circumferential involvement, curative resection, histologic type, and histologic grade were evaluated.

### Pre-operative preparation and surgical approaches

In our hospital, multispecialty approaches with careful preoperative evaluations are performed for these patients. Several preoperative considerations are listed as follows:

1. Nutrition status evaluation with supplement if indicated.
2. Maging studies (CT scan or magnetic resonance imaging) to determine the tumor location and size, the relationship of the tumor to the adjacent structures, resection margin, and surgical approach.
3. Preoperative selective coil arterial embolization can be considered for highly vascularized tumors to reduce blood loss and improving clarity of surgical field.
4. Ureteric stents before tumor manipulation may aid identification of the ureters in cases of accidental injury.
5. Prepare equipments necessary for massive transfusion intraoperatively.
6. Ensure appropriate positioning of the patient according to the planned surgical approach while protecting pressure points to prevent pressure injury to the soft tissues and the nerves.

The selection of the surgical procedure depends on multiple factors. These factors should be considered in the qualification process, which include tumor size, its precise location relative to the adjacent organs, and degree of vascularization. Special attention should be paid to the relationship between the tumor and S3 vertebra, the location relative to the rectum,

and regional vascular plexuses and nerves.

### Statistical analysis

The collected data were analyzed using IBM SPSS Statistics for Macintosh, version 26.0 (Armonk, NY: IBM Corp.). Student's t-test and chi-square test were used to analyze continuous and nominal variables, respectively. Statistical significance was set at  $p < 0.05$ .

## Result

Thirty-six patients who underwent surgical intervention for presacral tumors were included. Of these patients, 13 men and 23 women. The mean age was 60 years (range, 29-84 years) (Table 1). These patients were admitted to three units, including the orthopedics, neurosurgery, and colorectal surgery departments of the Chang Gung Memorial Hospital, with various chief complaints. The most common symptoms were neurological or orthopedic symptoms including low back pain or sciatica (50%), followed by gastrointestinal symptoms (27.8%) (Table 1). All patients under-

went at least one imaging study (a CT scan, MRI, or transrectal ultrasound). One-fourth of the patients (9/36) underwent preoperative biopsy.

Thirty-one patients (86.1%) underwent curative resections (Table 2). Two patients underwent pure laparoscopic resection. The average operation time was 330 min. There were 14 cases (38.9%) in the com-

**Table 2.** Surgical records, morbidities, and pathology

Surgical methods	
Curative resection	33 (91.7%)
Palliative resection	3 (8.3%)
Surgical approach	
Anterior	14 (38.9%)
Posterior	8 (22.2%)
Combined	14 (38.9%)
Surgical time (min)	330 ± 28 (63-755)
Blood loss (mL)	1496 ± 512 (10-18100)
Tumor size (mm)	82.6 ± 6.6 (30-250)
Length of hospital stay (post-operative day)	10.6 ± 1.1 (3-30)
Morbidity	
Nil	15 (41.7%)
Wound infection	5 (13.9%)
Deep vein thrombosis	1 (2.8%)
Neurogenic bladder	10 (27.8%)
Drop foot	2 (5.6%)
Sciatica	2 (5.6%)
Fecal incontinence	1 (2.8%)
Paresthesia	3 (8.3%)
Enterocutaneous fistula	1 (2.8%)
Pathology	
Schwannoma	7 (19.4%)
Chordoma	14 (38.9%)
Teratoma	7 (19.4%)
Giant cell tumor of bone	2 (5.6%)
Epidermoid cyst	1 (2.8%)
Chondrosarcoma	1 (2.8%)
Metastatic adenocarcinoma from lung	1 (2.8%)
Cattleman disease	1 (2.8%)
Tailgut cyst	1 (2.8%)
Plasma cell myeloma	1 (2.8%)
Upper border (via image)	
L5	2 (5.6%)
S1	10 (27.8%)
S2	15 (41.7%)
S3	6 (16.7%)
S4	2 (5.8%)
Coccyx	1 (2.8%)
Recurrence	5 (13.9%)

**Table 1.** Demographics and clinical profiles

Age (years)	60 ± 2.37 (29-84)
Gender	
Male	13 (35.9%)
Female	23 (64.1%)
Division	
Orthopedics	19 (52.8%)
Neurosurgery	4 (11.1%)
Colorectal surgery	13 (36.1%)
Chief complaint	
Asymptomatic/incidental finding	7 (19.4%)
GI (anal pain, constipation, abdominal discomfort)	10 (27.8%)
Urologic symptoms	1 (2.8%)
Neurologic/orthopedic symptoms (low back pain, sciatica)	18 (50.0%)
Image	
CT	10 (27.8%)
MRI	11 (30.6%)
CT + MRI	14 (38.9%)
MRI + rectal ultrasound	1 (2.8%)
Pre-operative biopsy	9 (25%)

bined anterior and posterior approach group, 8 (22.2%) in the posterior approach group, and the remaining 14 were from the anterior approach group. The estimated average blood loss was approximately 1500 mL, ranging from 10 to 18 100 mL. The mean length of the postoperative hospital stay was 10 days. The most common diagnosis was chordoma, followed by schwannomas and teratomas.

Postoperative complications were up to 58.3% (21/36) of the patients. After surgery, the most common immediate complication was a neurogenic bladder, followed by surgical wound infections. Neurogenic bladder developed postoperatively in 10 patients, two of whom underwent further cystostomy to resolve urinary retention. Surgical wound infection occurred in 5 patients (5/36; 13.9%). Of these patients, 2 required additional surgical debridement. Of the 36 patients who underwent presacral tumor resection, 5 experienced tumor recurrences (3 chordomas, 1 chondrosarcoma, and 1 schwannoma) during follow-up.

Postoperative morbidity was higher in patients with tumors above the S3 vertebra than in those with tumors below the S3 vertebra (66.7% vs. 11.1%,  $p = 0.004$ , Table 3). The posterior surgical approach was more frequently employed in patients with tumors below the S3 vertebra (6/9, 66.7%) than in those with tumors above S3 (2/27, 7.4%) ( $p = 0.001$ ). Owing to more postoperative complications, the length of hospital stays in the above S3 group was longer than that

in the below S3 group (12.0 days vs. 6.7 days,  $p = 0.037$ ). There were no statistically significant differences among the other variables, including age, operation time, blood loss, tumor size, and diagnosis.

Among 10 patients who has neurogenic bladder, two of them received cystostomy. One patient who had complication of enterocutaneous fistula received diverting colostomy. In addition, two of five patients with surgical site infection undergone surgical debridement.

Five patients (13.9%) had recurrence. They were chordoma (3 cases) chondrosarcoma (1 case) and schwannoma (1 case).

## Discussion

In this study, 36 patients diagnosed with presacral tumors, either asymptomatic or symptomatic, underwent surgical resection. Chordoma is the most common presacral tumor diagnosis, followed by schwannomas and teratomas. Neurogenic bladder is the most common postoperative morbidity, followed by surgical wound infection. The incidence of postoperative complications in the above S3 group was higher than that in the below S3 group. A posterior approach strategy was more commonly used in the group with a presacral tumor where the upper border was below the S3 vertebra level.

Presacral tumors are a heterogeneous group of lesions with diverse histology, classified as benign or malignant, and congenital or acquired.<sup>2,3</sup> These heterogeneous tumors are challenging to categorize because the tissue covering the presacral region is derived from the embryonic stage. Inconsistent with the literature, our most common diagnosis was chordoma, followed by teratoma and schwannoma.<sup>9-11</sup> Of the tumors in this study, 34% (10/29) were malignant tumors, which is slightly varied from the findings of other studies.<sup>9</sup> Since we are a referral center, this difference may be related to the type of patients referred and the greater complexities of their conditions. In addition, we only collected patients with presacral tumor receiving combined surgery with colorectal surgeon which might lead to this different result.

**Table 3.** Comparison between tumors above and below S3 vertebra

	Above S3 (n = 27)	Below S3 (n = 9)	<i>P</i>
Age (years)	60.7 ± 12.6	58.0 ± 18.9	0.069
Morbidity	18 (66.7%)	1 (11.1%)	0.004
Surgical approach			0.001
Anterior	13	1	
Posterior	2	6	
Combined	12	2	
Surgical time (min)	366.1 ± 168.7	223.3 ± 117.3	0.142
Blood loss (mL)	1856.1 ± 3471.4	416.7 ± 590.7	0.194
Tumor size (mm)	93.1 ± 39.9	51.1 ± 13.4	0.108
Length of stay (d)	12.0 ± 6.6	6.7 ± 3.4	0.037

POD = postoperative day.

In our study, tumors above the S3 vertebra were primarily resected using an anterior or combined approach, while a posterior approach was used for tumors below the S3 vertebra. At our institute, these complicated tumors are usually resected by two or more diverse specialty surgeons including orthopedic surgeons, neurosurgeons, and colorectal surgeons. Orthopedics and neurosurgeons are generally responsible for the primary tumor resection, including bone resection, if indicated. In contrast, colorectal surgeons are responsible for tumor mobilization, including dissection of the tumor from the colorectum and the adjacent adhered organs, such as the ureter, iliac vessels, and the sacral nerve. Most of these adjacent structures can be technically manipulated, including resection. Nevertheless, significant morbidity can occur, especially in fecal and urinary functions. In this series, 10 patients (27.8%) experienced a neurogenic bladder, and 2 of them underwent cystostomy due to refractory medical treatment and repeated urinary tract infections after extensive bony resection and bilateral nerve injury. Furthermore, one patient (2.8%) developed fecal incontinence.

Although unilateral resection of the sacral nerve roots does not completely impair fecal and urinary functions,<sup>6,9</sup> these complications may occur if both S3 nerve roots are damaged. Extensive sacrotomy is associated with significant morbidity and dysfunction. In addition, proctectomy with or without colostomy may compromise the patient's quality of life.

Therefore, a comprehensive assessment of the upper border of the presacral tumor and the adjacent organ involvement before surgery is necessary. Detailed preoperative investigations can be performed given the improved imaging modalities, including CT and MRI. To minimize disparities in outcome expectations after surgery, it is essential to discuss the details with the patients and their families, including the benefits and risks of extended resection.

This study had several limitations. First, the use of a retrospective study based on medical records may have led to biases. Second, the operative approach varied among surgeons with different specialties. The surgeon was the most influential decision-maker in the final surgical approach after discussions with the

patients and their families. In addition, surgeon's technique may also play important role in patient's outcome. Third, this study only included patients who had undergone surgical resection, which is non-representative of the actual incidence and the general characteristics of presacral tumors. Fourth, we collected patients received combined operation with colorectal surgeon only which means some patients with presacral tumor receiving surgery in orthopedic and neurosurgery divisions were not collected.

## Conclusion

Presacral tumors are rare, and often without specific clinical symptoms. The location of these tumors makes diagnosis difficult and often delayed. Preoperative imaging studies, such as computed tomography and magnetic resonance imaging, are the key to surgical planning. Surgery is the primary treatment for this disease, with a risk of postoperative sequelae. Tumor location above the S3 vertebra is the main risk factor for postoperative complications.

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原 著

## 骶前腫瘤的手術治療：單一醫學中心 二十年的經驗

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**背景** 薦椎前腫瘤是一種相對罕見的病灶。對於這種疾病的適當手術方式仍然沒有非常明確的治療指引。

**目的** 本研究旨在調查接受手術的骶前腫瘤患者並回顧這種罕見疾病。

**材料和方法** 回顧性研究於 2000 年 1 月至 2022 年 12 月期間在林口長庚醫院接受薦椎前腫瘤手術的患者。

**結果** 共收集 36 名患者。13 名患者為男性，23 名患者為女性。最常見的主訴是神經性或骨科症狀，如腰痛或坐骨神經痛 (50%)，其次是胃腸道疾病 (27.8%)。共有 2 例為純腹腔鏡切除手術。手術方面，14 例 (38.9%) 患者採用合併經腹部及會陰路徑，8 例 (22.2%) 經會陰路徑，14 例 (38.9%) 經腹部路徑。平均失血量約為 1500 毫升。術後平均住院時間為 10 天。四分之一的患者術後出現神經性膀胱。最常見的病理診斷是脊索瘤 (14 例，38.9%)。位於第三薦椎骨以上的腫瘤比，第三薦椎骨以下的腫瘤有更多的術後併發症 (66.7% 對 11.1%， $p = 0.004$ )。

**結論** 腫瘤上緣位於第三薦椎骨以上是術後併發症的重要危險因素。詳細的術前計劃和縝密的手術策略對於較佳的結果是必要的。

**關鍵詞** 骶前腫瘤、直腸後腫瘤。