The Treatment Efficacy of Transparasacral Approach for Presacral Tumor: A Single Center Experience

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Abstract

Background

Presacral tumors, also known as retrorectal tumors, locate in the presacral space and are clinically rare and the approaches for the diagnosis and treatment of presacral tumors are still deficient. The four accepted surgical approaches for presacral tumors include transabdominal approach, sacrococcygeal approach, perineal approach, and combined approach. This study aims to evaluate the treatment efficacy of transparasacral approach for presacral tumors.

Methods

23 patients (7 males and 16 females) who are diagnosed with presacral tumors and receive surgery in transsacral approach at our department were chosen. The gender, age, body mass index (BMI), and clinical symptoms were recorded. The clinical data, postoperative complications, and short and long-term rehabilitation data were recorded and analyzed.

Conclusions

The surgery was performed smoothly and the patients recovered well without signs of intestinal fistula, abdominal and pelvic infection, lung infection, organ dysfunction, or organ failure. The incision edema occurred in only one patient. In terms of the histopathological types, presacral tumors can be classified into tailgut cyst ($n = 10$), epidermoid cyst ($n = 5$), teratoma ($n = 3$), and lipoma ($n = 10$). One patient diagnosed with tailgut cyst was accompanied by adenocarcinoma transition. Tumor recurrence didn't occur in any patients. In addition, no patients complained about dysuria, sacrococcygeal sensory disturbance, or sacrococcygeal and perineal discomfort. Surgical resection with transparasacral approach is effective for the treatment of presacral tumors with the advantages of high safety, good treatment efficacy, few complications, good oncological prognosis, and fast recovery.

Introduction

Presacral tumor, also known as retrorectal tumor, grows in the presacral space between the rectum and the sacrum and is clinically rare (1). Only one presacral tumor patient among 40,000 to 60,000 patients needs surgical treatment (2). Presacral tumors are characterized by insidious onset, complicated pathological types, large size, and surgery difficulty (3, 4). The approaches for the diagnosis and treatment of the presacral tumors are still deficient.

The presacral space is a latent space deep in the pelvic cavity (Fig. 1). The boundary of presacral space consists of peritoneal reflex, levator ani muscle, rectal fascia, sacrum, bilateral ureters, bilateral iliac blood vessels, and bilateral sacral nerve roots (5). The presacral space contains a variety of tissues, including

the tail bowel, notochord, and other embryonic residual tissues, which may develop into presacral tumors. In terms of the pathological types, presacral tumors can be divided into congenital tumors accounting for about 65%, neurogenic tumors, bone-derived tumors, and other cell-derived tumors (5, 6).

Presacral tumors are asymptomatic in the early stage due to the small size, which may delay the diagnosis and even lead to misdiagnosis. When the volume increases to a certain extent, local compression or infection occurs (5). The digital rectal examination, imaging examination, and colonoscopy are vital for the diagnosis of presacral tumors (7). However, whether to perform the biopsy pathology is controversial as non-surgical treatments are more suitable for some malignant tumors, and the pathological results can be adjusted in time before surgery. Besides, the biopsy may lead to the spread of malignant tumor tissues and local infection.

Surgery is vital for the treatment of presacral tumors and the four accepted surgical approaches are transabdominal approach, sacrococcygeal approach, perineal approach, and combined approach (5). The transabdominal approach can be divided into laparotomy and laparoscopic approaches and is applied when the grade of the tumors is above S3 (5). The sacrococcygeal approach includes the sacrococcygeal midline approach and transparasacral approach and is often applied to the treatment of tumors of grade S3 or below. The perineal approach is mainly applied for the treatment of rectal adenoma or tumors in the retro-rectal space (8). The Sacrococcygeal approach can shorten the distance to the lesion, expose the tumor, remove the coccyx and protect the surrounding tissues and organs (9). The transparasacral approach can adjust the incision position based on the location of the tumor to reduce trauma, shorten incisions, and reduce operation time.

This study evaluates the treatment efficacy of transparasacral approach for presacral tumors with 23 patients undergoing transparasacral approach treatment in our department.

Materials And Methods

General data

This observational and prospective study was approved by the Ethics Committee and Institutional Review Board of our institution. All methods were performed in accordance with the relevant guidelines and regulations. 23 patients (7 males and 16 females) who were diagnosed with presacral tumors at the Department of General Surgery of our hospital from July 2016 to December 2020 and received transsacral approach surgery were chosen. The gender, age, body mass index (BMI), and clinical symptoms were recorded.

Inclusion criteria

The patients who were diagnosed with presacral tumors via imaging examinations and received surgical treatment via parasacral approach without undergoing other surgical treatments at the same time.

Exclusion criteria

(1) The patients with malignant tumors metastasizing from other tissues and organs to the presacral space. (2) Patients who cannot tolerate anesthesia and surgery due to heart, lung, and other related organ dysfunction, or coagulopathy. (3) Patients undergoing other surgeries at the same time. (4) Patients with incomplete clinical data.

Preoperative preparation

(1) All the patients received the rectal digital examination. (2) Computed tomography (CT), magnetic resonance imaging (MRI), and/or ultrasound were performed to confirm the basic characteristics of the presacral tumors. (3) Biopsy pathology was not performed. (4) Colonoscopy was performed to exclude tumors derived from intestinal tissues. (5) General biochemical examinations were performed before surgery.

Surgery procedure

All patients underwent surgeries conducted by the same medical team via the transparasacral approach. The detailed surgery procedure was shown in Figure 2.

(1) Epidural anesthesia combined with subarachnoid intubation was performed with patients in lateral position or folding-knife position. (2) An incision with a length of 8-10 cm along the sacrum and coccyx was made. The position and length of the incision can be adjusted according to the preoperative imaging examination. (3) The skin, subcutaneous tissue, deep fascia, and part of the gluteus maximus muscle were cut layer by layer until the lateral margin of the sacrococcygeal joint was reached. Part of the anal tail ligament was cut off, and if necessary, part of the coccyx and even the sacrum were removed. The sacrum and adjacent nerves should be preserved during the treatment of tumors with the grade of S3 or above. (4) The retrorectal and presacral spaces were exposed. The tumors should be resected carefully. When resecting the upper edge of the tumors, ligation of blood vessels should be performed if necessary, to avoid bleeding of the presacral venous plexus. (5) During the surgery, the posterior wall of the rectum, rectal pubic muscles, internal and external rectal sphincter, and ureter should be protected and the protection of the posterior wall of the rectum is performed based on rectal digital examination. For female patients, the vagina and uterus should also be protected. (6) If the tumors are cystic and capsular, and the large volume makes the separation difficult, it is feasible to decompress the cysts by draining out the liquid contents or pinching out the solid contents. (7) When the tumors and the margin are completely resected, the wound should be electrocoagulated to stop the bleeding. Rapid pathology should be performed if necessary. A drainage tube should be placed in the presacral space.

Clinical observation

All patients were given anti-inflammatories and fluid replacement treatment. The clinical data were recorded, including operation time, amount of bleeding, time of indwelling drainage tube, whether to remove coccyx, incision length, the maximum diameter of tumor, whether the resection is complete, and pathological type. Postoperative complications were also noted, such as mortality, intestinal fistula,

abdominal and pelvic infection, lung infection, organ dysfunction, and organ failure, incision edema, poor incision healing, *etc.* In addition, the short and long-term rehabilitation data were recorded. The short-term data include starting time of drinking water, eating liquid diet and eating half-liquid diet; first ambulation time; time to pull out the drainage tube; first urination time; first defecation time; hospital stay; return to work time, *etc.* Long-term rehabilitation data include the incidence of tumor recurrence, difficulty urinating, difficulty defecating, and sensory disturbances, as well as discomfort in the sacral and perineum.

Statistical analysis

The statistical analysis was performed by SPSS 22.0. The data were presented as mean $\pm$ SD.

Results

Based on the inclusion and exclusion criteria, 23 patients, including 7 male (30.43%) and 16 female (69.57%) patients were chosen for this study (Fig. 3). The follow-up time was 4–59 months.

The patients were 42.87 ± 10.43 years old with the youngest being 29 years old and the oldest being 76 years old. The BMI of these patients was $23.06 \pm 2.51 \text{ kg/m}^2$.

In our study, 19 cases (82.61%) were asymptomatic, two cases (8.70%) had an anal bulge, one case (4.39%) had lower abdominal pain, and one case had sacral skin bulge. Two patients had a history of abdominal pelvic surgery, and one had undergone presacral cyst resection through the abdominal approach. The other had undergone hysteromyomectomy. All patients underwent rectal digital examination and all presacral tumors could be touched. CT and MRI examinations were performed for all the patients. All the patients underwent color Doppler ultrasound examination: 15 patients underwent abdominal ultrasound examination, 4 patients underwent vaginal ultrasound examination, and 4 patients underwent colorectal ultrasound examination. Presacral tumors were found in all imaging examinations. Colonoscopy was performed for all patients and the compression of the rectum was found while the primary colorectal tumor was not found. The upper edges of the tumors of two patients were above grade S3.

All surgical procedures were performed smoothly. The intraoperative clinical data are listed in Table 1.

Table 1
Intraoperative clinical data of patients with presacral tumors.

Intraoperative clinical data	
Operation time (min)	80.43 ± 15.80
Bleeding (mL)	73.48 ± 31.14
Intraoperative blood transfusion (mL)	0
Incision length (cm)	8.57 ± 1.75
Maximum of tumor (cm)	10.98 ± 1.65
Complete resection (%)	100
Coccyx resection (cm)	100

All patients recovered well after surgery without signs of intestinal fistula, abdominal and pelvic infection, lung infection, organ dysfunction, or organ failure. The incision edema occurred in only one patient.

The histopathological types of presacral tumors are listed in Table 2. One patient diagnosed with tailgut cyst was accompanied by adenocarcinoma transition.

Table 2
Histopathological types of presacral tumors.

Histopathological types	Tailgut cyst	Epidermoid cyst	Teratoma	Lipoma
Number	10	5	3	5
Percentage (%)	43.48%	21.74%	13.04%	21.74%

Table 3 provides the short-term rehabilitation data. Until May 2021, tumor recurrence didn't occur in any patients. In addition, no patients complained about dysuria and dysuria, sacrococcygeal sensory disturbance, or sacrococcygeal and perineal discomfort.

Table 3
Short-term rehabilitation data.

Rehabilitation data (day)	
Time to get out of bed	2.78 ± 0.74
Analgesics administration time	4.78 ± 1.27
Time to pull out drainage tube	7.12 ± 1.23
Time to recover drinking water	2.78 ± 0.79
Full-flow food recovery time	3.91 ± 0.73
Half-flow food recovery time	4.78 ± 0.95
Time to resume urination	2.74 ± 0.75
First postoperative defecation time	3.39 ± 0.58
Length of hospital stay	14.17 ± 2.53
Postoperative hospital stay	10.17 ± 2.53
Return to work time	11.91 ± 2.83

Discussion

Presacral space is deep and contains a variety of tissue structures, such as loose connective tissue, sacral plexus branches, sympathetic nervous system, arteriovenous system, tail bowel, notochord, and embryonic residual tissue (4, 10, 11), which may develop into presacral tumors. Early diagnosis and treatment of presacral tumors are difficult, and misdiagnosis is easy to occur. The abdominal and pelvic symptoms of patients with presacral tumors are not obvious, and most tumors are found by CT and other imaging examinations. The volume of the tumor is often very large at the time of discovery, which adds surgery difficulty.

Only one presacral tumor patient among 40,000 to 60,000 patients needs surgical treatment (2). The incidence of presacral tumors of the female is 2–15 times larger than that of the male (5). In this study, the incidence rate of the female is 69.57%, which confirms the finding in the previous study. Two patients had a surgery history of abdominal or pelvic diseases, which didn't increase the difficulty of the resection of presacral tumors.

The complexity of tissues in presacral space leads to the diversity of pathological types of presacral tumors. The classification criteria for the pathological types of presacral tumors have not been unified. The pathological characteristics of presacral tumors in children and adults are different. In addition, the researchers haven't reached an agreement on whether malignant sacral tumors or neurogenic tumors that invade the presacral space should be included (10). The classification system proposed by Uhlig and

Johnson contains at least 25 different histological types that have been widely used (12). The most common one is developmental cysts, ranging from that with a diameter of a few centimeters to lesions that fill the pelvis (13). Due to the complexity of the contents of the presacral space and the embryonic residual tissues in the presacral space, presacral tumors always originate from a variety of organs and tissues, and about 60% are congenital residual tissues (1). It is reported that 65% of the presacral tumors are congenital, 12% are neurogenic, 11% are osteogenic, and 12% are of other cellular origins (14). In this study, there were 18 congenital presacral tumors, accounting for 78.26%. About 45%-50% of presacral tumors are malignant or accompanied by malignant transformation (14, 15), and the malignant rate of solid tumors (60%) is higher than that of cystic tumors (10%) (16). Tailgut cysts with malignant transformation are very rare and usually belong to the pathological types of transitional cell carcinoma, carcinoid, and adenocarcinoma (17, 18). In our study, there was only one case with a tailgut cyst accompanied by adenocarcinoma transition. Malignant tumors that metastasize to the presacral space are not included in this study, because they are small, scattered, and independent, and the research of them belongs to another specialized field. Most primary presacral malignancies are insensitive to radiotherapy or chemotherapy (5, 19), which can only reduce the pain of patients but cannot resect the tumors completely. However, specific radiotherapy or chemotherapy based on the characteristics of metastatic tumors in the presacral space can be applied.

In this study, 19 patients were asymptomatic, accounting for 82.61%, which may be attributed to that the initial tumor is small and is located deeply in the limited presacral space, which restricts the growth of the tumor. However, as the tumor grows, local compression such as lower abdominal, pelvic, or sacral pain, constipation (20, 21), and anal bulge (22) and infection may appear. The large presacral tumor can also lead to intestinal obstruction (23), irritable bowel syndrome (24), gait abnormality (25). If the tumor capsule ruptures, the outflow of the tumor contents can also lead to infection, such as recurrent pilonidal hairs (26), pelvic abscesses (27), and perianal abscesses (28). Furthermore, in our study, one case had a sacral skin bulge, which hasn't been reported in previous studies. It can be concluded that most patients with presacral tumors are asymptomatic and a small number of patients only present non-specific symptoms. The low incidence of the disease and the limited medical experience often result in delayed diagnosis and treatment.

Rectal digital examination and imaging examination are vital for the diagnosis of presacral tumors. Digital rectal examination is the most efficient, simple, and economical clinical method (10), and most presacral tumors can be touched by digital rectal examination (29). Digital rectal examination can not only obtain the basic information of tumors at the early stage, such as the location, size, texture, and adjacency but also guide the selection of surgical methods (5). If the upper edge of the presacral tumor can be touched by rectal digital examination, the tumor is very likely to be resected with the sacrococcygeal approach.

Imaging examinations of presacral tumors mainly include CT, MRI, abdominal color Doppler ultrasound, intrarectal ultrasound, and female vaginal color Doppler ultrasound examination. Abdominal color Doppler ultrasound is an economical and practical examination that can provide a high positive

diagnostic rate (18). Intrarectal ultrasound, which can indicate whether the tumor is cystic, substantial, or mixed, and can be used to explore the tumor location and adjacent relationship is important for the diagnosis, intraoperative guidance, and postoperative monitoring. Intrarectal ultrasound should better be combined with colonoscopy. The diagnostic positive rate of Female transvaginal color Doppler ultrasound should be further studied. CT and MRI can provide basic information of presacral tumors. Compared with MRI, CT is more sensitive to calcification of tumor tissues (5). The three-dimensional reconstruction of CT can intuitively show whether the adjacent tissue structure is involved, and the enhanced CT scan shows the blood supply of the tumor. MRI is more sensitive to adipose, necrosis, and bleeding and plays an important role in evaluating the relationship between tumor and sacrum, judging whether the sacrum needs to be removed, and determining the extent of resection (5, 30). It is reported that the sensitivity and specificity of imaging examinations for presacral tumors are 83% and 81%, respectively (31). Preoperative colonoscopy has the advantage of excluding primary colorectal tumors and determining the rectal involvement. All these examinations provide important information for preoperative evaluation of presacral tumors.

Whether to perform a preoperative biopsy for presacral tumors is controversial. Some researchers hold the opinion that histopathological biopsy is necessary to distinguish benign and malignant presacral tumors (32). In addition, based on the histopathological types, the individualized cancer treatment plan can be better applied. In a study performed by Messick *et al.*, the treatment plan of 3 cases changes from surgical treatment to chemotherapy or radiotherapy (32). However, due to tumor seeding, especially that of malignant presacral tumors, some doctors are reluctant to perform a pathological biopsy for presacral tumors. Furthermore, when performing a biopsy, it is easy to damage surrounding tissues, blood vessels, and nerves (5). For different types of presacral tumors, biopsy puncture may lead to different problems. For example, cystic tumors and inflammatory masses may rupture due to puncture, leading to infection. Another example is that neurogenic tumors may lead to nerve damage due to the puncture (5). From the perspective of our clinical team, whether to perform preoperative biopsy should be determined in specific situations. First, in most cases, biopsy is not recommended. Second, if the tumor is large and invades nearby tissues, which adds the difficulty in complete resection, histopathological biopsy can be applied after fully consideration. Third, transrectal or transvaginal biopsy should be prohibited, and transcutaneous biopsy (transabdominal, transsacral, transperineal, *etc.*) under color ultrasound or CT guidance is recommended as the biopsy tract can be resected while removing the tumor.

In this study, 23 patients underwent surgeries with the transparasacral approach. Compared with the transparasacral approach, the other three accepted surgical approaches mentioned above have their drawbacks.

Transabdominal surgery includes laparotomy and laparoscopy surgery. The disadvantages of laparotomy for the treatment of presacral tumors include: (1) The surgical path to the lesion is the longest because surgeons need to pass through the abdomen before entering the pelvic cavity. (2) The incision should be long enough to ensure a good surgical view. (3) It is difficult to get the caudal side of the tumor in the narrow anterior sacral space deep in the pelvic cavity. (4) The coccyx cannot be resected,

which increases tumor recurrence rate. (5) If the patients have a history of abdominal or pelvic surgery, the risk and difficulty of surgery will increase. It is reported that laparoscopic surgery is effective for the treatment of presacral tumors. However, laparoscopic surgery still has some drawbacks: (1) The surgical path is still the longest. (2) The coccyx cannot be removed. (3) Abdominal or pelvic surgical history increases the surgery difficulty. (4) High medical expenses.

The perineal approach is only suitable for a small number of patients, especially those with small presacral tumors (32). The perineal approach has obvious disadvantages for the treatment of presacral tumors, including: (1) The surgical field is small and is not suitable to resect tumors with large volumes. However, the common presacral tumors are large, which makes adds difficulty to surgery. (2) The incision is close to the anus, which is not beneficial for wound healing. (3) The application is limited as the tumor cannot be too far away from the perineal incision. (4) The coccyx cannot be resected.

The combined approach has the disadvantages of the long operation time, a large number of incisions, the need to change the patient's position, and a large wound surface, which will inevitably lead to poor applicability.

The advantages of the transparasacral approach include: (1) The surgical path is short, which decreases the surgery difficulty. Presacral tumors are large at the time of diagnosis, but are usually lower than grade S2 and grow toward the tip of the coccyx (33). Through the skin, subcutaneous tissue, deep fascia, and part of the gluteus maximus, surgeons can expose the tumor, which can minimize the secondary damage to the surrounding tissues and organs. (2) The incision is short and the tumor can be better exposed. Based on the preoperative imaging examinations, the position of the incision can be adjusted to make the distance from the incision to the tumor the closest. In this situation, the surgical field is wide, the operation space is large, and the surgery risk is small. All tumors in this study were successfully removed through the transparasacral approach. Tumors with a diameter of less than 10 cm can be fully exposed with the transparasacral incision. Even polycystic cysts can be exposed and completely removed after being fully decompressed. (3) The coccyx can be easily removed. The resection of the coccyx or even part of the sacrum can well expose the tumor, reduce the tumor recurrence (34) and help achieve R0 resection. (4) The surgery-related risk and surgery difficulty for patients with abdominal or pelvic surgery history will be decreases. In our study, two patients had an abdominal or pelvic surgery history, but the surgery was finished smoothly with the transparasacral approach. (5) In our study, the upper edges of the tumors of two patients were higher than grade S3 and the tumors were also removed successfully, indicating that transparasacral approach may also be applied for presacral tumors higher than grade S3. (6) Because of the small incision and few injuries to adjacent tissues, patients who underwent transparasacral approach always recover fast. (7) Transparasacral approach provides a satisfactory treatment efficacy for presacral tumors. In our study, during the follow-up time ranging from 4 to 59 months, no tumor recurrence has been found. In addition, the urination function, defecation function, and skin sensory function were not being affected, and there was no discomfort in the sacral tail, perineum, *etc.*

The surgical skills of the transparasacral approach for the treatment of presacral tumors are concluded here. (1) During the operation, surgeons should pay attention to the important tissue in the operation field, such as pelvic nerve plexus, blood vessels in the iliac fossa, ureter, anterior sacral venous plexus, and rectum. Digital rectal examination can help preserve the rectum. (2) As the presacral tumors may not be separated from the rectum or vagina, intestinal preparation is necessary before surgery. For most benign cystic lesions that compress the rectum, the tumors can be removed successfully without rectum resection. However, for malignant tumors, the resection of part of the intestine may be needed. (3) The principle of no tumors should be strictly followed. Squeezing of the tumor should be prohibited and the tumor capsule should be better preserved. If the cyst is ruptured during the operation, the surgical field should be washed repeatedly with saline to avoid contamination. (4) If the lesion is in the stage of infection and edema, anti-infective and anti-inflammation treatment should be performed. (5) Whether to perform the intraoperative decompression should be based on the preoperative evaluation. If the tumor is cystic, intraoperative decompression can be performed. If the tumor is mixed cystic or solid and cannot be excluded from malignant lesions, intraoperative decompression should be inhibited. (6) Ligation of blood vessels should stay away from the tumor to avoid surface bleeding of the tumor. (7) The tumor should not be separated near the sacrum to avoid the bleeding of the anterior sacral plexus. (8) The Henson nodule on the coccyx is the site where multifunctional cells are gathering and must be removed during the operation to prevent tumor recurrence (35). It can be said that resection of the coccyx during the operation not only broadens the surgical field but also meets the needs of radical resection. (9) Drainage with negative pressure should be placed in the presacral cavity after tumor resection.

In conclusion, the surgery with the transparasacral approach is effective for the treatment of presacral tumors with the advantages of high safety, good treatment efficacy, few complications, good oncological prognosis, and fast recovery.

Abbreviations

BMI

body mass index; MRI:magnetic resonance imaging.

Declarations

Acknowledgement

Not applicable.

Authors' contributions

Study conception and design: WM and LGL; Acquisition of data: LGL; Data analysis: ZLX and HHY; Drafting the manuscript: WM; Critical revision: LJN.

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Data availability

Request with reasonable grounds from corresponding author.

Ethics approval

This study was approved by the Ethics Committee and Institutional Review Board of the Second Hospital of Jilin University, Changchun, China.

Consent to participate

Informed consent was obtained from all patients.

Consent for publication

All authors consent to publish this work.

Competing interests

The authors declare no competing interests

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Figures

Figure 1

Illustration of presacral space.

Figure 2

Surgery procedures. (A) Transsacral incision. (B) Excision of the coccyx. (C) Separating the tumor capsule. (D) Dissecting the upper edge of the tumor. (E) Completely removing the tumor. (F) Placement of the drainage tube.

Figure 3

Patients included in our department from July 2016 to December 2020.