



Decision Making for Surgical Approaches to Resection of Benign Presacral Tumors in Adult Women: A Case Series

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Abstract

Study Objective: To explore decision making strategies for the surgical approach to resection of benign presacral tumors of different sizes and locations.

Design: Retrospective case series.

Setting: An academic tertiary care university hospital in Guangzhou, China.

Patients: Four adult female patients with benign presacral tumors who were admitted to our department for surgical treatment of benign presacral tumors between 2015 to 2020.

Interventions: Minimally invasive surgical resection of presacral tumor.

Measurements and Main Results: The incidence of benign presacral tumors is relatively low in adults and is much higher in women than in men. However, there is no standard surgical approach for resection of presacral tumors in women. We report 4 women in which benign presacral tumors were removed via a transvaginal, laparoscopic, and combined laparoscopic-vaginal approach. The surgeries were all carried out successfully, and each patient underwent appropriate post-operative follow up without tumor recurrence.

Conclusion: Different approaches to the surgical resection of benign presacral tumors exist, and decision making strategies are necessary to determine the most appropriate intervention in various patients. The transvaginal approach is an uncommon but feasible and safe approach for such resection in adult females.

Keywords: Benign presacral tumor; Transvaginal surgery; Adult women

Abbreviations

CT: Computed Tomography; S1: The First Sacrum; S3: The Third Sacrum; S4: The Fourth Sacrum; S5: The Fifth Sacrum; MRI: Magnetic Resonance Imaging

Introduction

The benign presacral tumor is a mass located in the retrorectal or presacral space, which is a space bound by the posterior rectum anteriorly, the sacrum and coccyx posteriorly, and the anococcygeal ligament inferiorly. According to the database of hospital admissions at the Mayo Clinic from 1922 to 1936, benign presacral tumors occurred in one in every 40,000 hospital admissions [1]. The incidence of such tumors was further estimated at 6.3 individuals per year based on the same database between 1960 and 1979 [2]. Therefore, the incidence of benign presacral tumors appears to be uncommon, but the true incidence is unknown as this information was derived from a single large medical center and may not be generalizable to the entire population [3].

During embryonic development, different layers of embryonic cells converge in presacral space. Therefore, various types of lesions may develop in this space. The presacral tumor can be broadly classified into developmental, neurogenic, osseous, and miscellaneous tumors [4]. Most of these tumors are benign developmental tumors, such as dermoid or epidermoid tumors [5].

Most patients who develop such tumors are female [6], and clinical symptoms are unspecific

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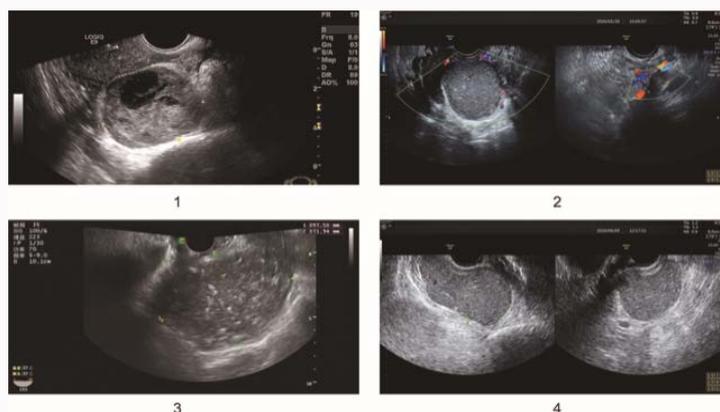


Figure 1: The benign presacral tumors of patients involved in Doppler ultrasound.

[5]. Nearly half of patients are diagnosed with presacral tumors incidentally through routine physical examination or during imaging studies. If patients become symptomatic, that may complain of sacro-perineal pain, constipation, or dysuria [7] due to compression of adjacent organs by the tumor. Once the diagnosis is confirmed, surgical removal is appropriate in order to prevent potential malignant transformation or secondary infection [8]. The anterior (abdominal) approach, the posterior (perineal) approach and the combined (anterior-posterior) approach are the most common described surgical approaches [9]. However, the transvaginal approach in adult women should also be considered [10,11].

Unfortunately, there is limited information describing how to perform a transvaginal approach in the resection of benign presacral tumor for female patients. In this study, we describe 4 cases of minimally invasive resection of benign presacral tumor performed by a gynecological oncologist through transvaginal surgery, vaginal assisted transabdominal laparoscopic surgeries and pure transabdominal laparoscopic surgery without postoperative complications. The aim of this study is to propose a strategy of surgical approach decision-making for tumor removal exclusively for adult women. This study has been approved by the Ethics Committee of Guangdong Provincial Hospital of Chinese Medicine.

Case Series

Case 1

A 43-year-old female patient, JZL, was admitted for evaluation of an asymptomatic pelvic mass. The inferior aspect of the tumor was not palpable & vaginal or rectal examination. Cancer biomarkers, including serum Carbohydrate Antigen 125 (CA-125), Carbohydrate Antigen 153 (CA-153), Human Epididymis Protein 4 (HE4), Neuron-Specific Enolase (NSE), Carcinoembryonic Antigen (CEA), and Alpha Fetoprotein (AFP), were within normal limits. Transvaginal Doppler Ultrasonography (Figure 1.1) demonstrated a tumor behind the rectum, measuring approximately 50 mm × 49 mm × 40 mm with scattered increased echogenicity and a thin and smooth wall. Blood flow to the mass was normal. On CT (Computed Tomography) (Figure 2.1), the apex of the tumor was at the same level as the superior articular facet of the first sacrum (S1), and its inferior aspect at the same level as the superior articular facet of S4. In the horizontal plane (Figure 3.1), the tumor was located to the right of the sacrum without adhesions.

At the time of laparoscopy, the whole tumor could be visualized

after sigmoid colon was mobilized (Figure 4.1). Three abdominal trocars were used at the time of laparoscopy. One 10 mm trocar was inserted through the umbilicus for the laparoscope; two 5 mm trocars were located in the right lower and the left lower quadrant with use of the fenestrated bipolar forceps and the ultrasonic scalpel. The patient was placed in a Trendelenburg position with a slight left tilt for the entirety of the surgery. The retro-rectal space was entered anterior to the tumor by the incising the medial side of the uterosacral ligament, and the presacral tumor was then enucleated off meticulously along the tumor wall. During this dissection, the surgeon needs to be aware of adjacent vital organs, such as the ureter, sacral nerve plexus, rectum, and blood vessels. Therefore, maintaining a clear surgical field is pivotal and excellent hemostasis is necessary for successful surgery.

The operation took 120 minutes to complete and estimated blood loss was 20 mL. The final pathological diagnosis of specimen was reported as schwannoma. The patient did not have any intra- or postoperative complications and was discharged on postoperative day 9. She denied immediate postoperative pain or discomfort, but longer term follow up was not possible as she has not re-presented to clinic.

Case 2

A 49-year-old female patient, HYM, was diagnosed incidentally with a presacral tumor during bimanual vaginal and rectal examination. The inferior aspect of the tumor was palpable 4 cm above the hymen, but the superior aspect of the tumor could not be reached on exam. She reported no complaints and was otherwise asymptomatic. Cancer biomarkers, including CA-125, CA-153, HE4, SCC, NSE, CEA and AFP, were within normal limits. Transvaginal Doppler ultrasonography (Figure 1.2), demonstrated a 51 mm × 44 mm × 47 mm mass with scattered hyperechoic material and a thin and smooth walled cyst without aberrant blood flow. CT (Figure 2.2) demonstrated that the superior aspect of tumor was at the same level as the superior articular facet of S4 and the inferior aspect at the same level as the tip of coccyx. In the horizontal plane (Figure 3.2), the tumor was located to the right of the sacrum without adhesions.

At time of laparoscopy, only a part of the mass could be seen (Figure 4.2). Access to the tumor was performed similarly to Case 1. However, because the inferior aspect of the tumor was much lower compared to that of the tumor in Case 1, an assistant surgeon was necessary to push the tumor transvaginally, which elevates the tumor and allows for better exposure after successful dissection of the superior aspect of the tumor from adjacent structures. This allows

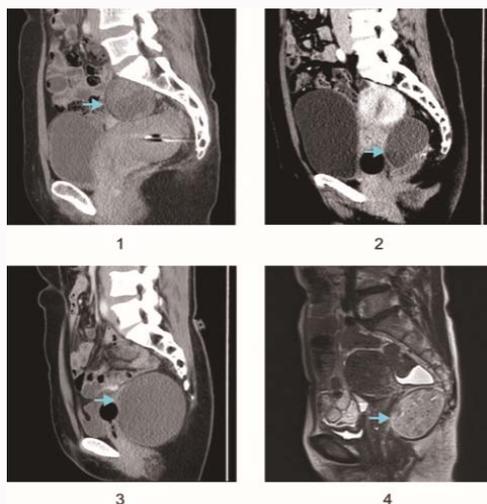


Figure 2: Sagittal images of benign presacral tumors on CT/MRI. The tumors are marked by the blue arrows.

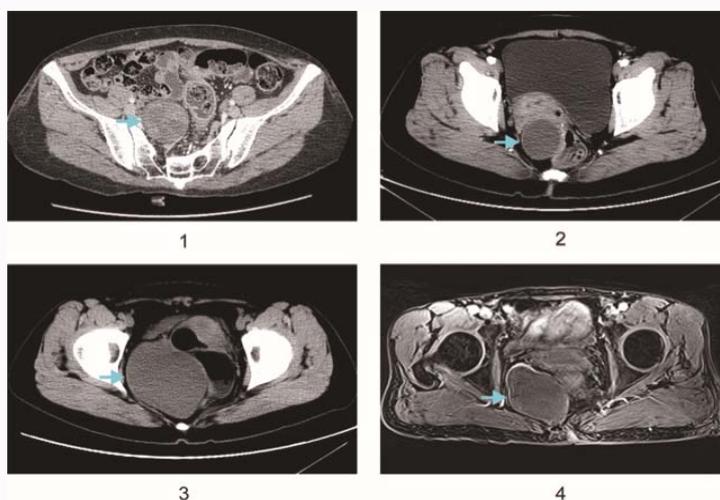


Figure 3: The horizontal images of benign presacral tumors on CT/MRI. The tumors are marked by the blue arrows.

for complete surgical resection of the tumor. The operation took 214 minutes to complete and estimated blood loss was 100 mL. The final pathological diagnosis of specimen was reported as epidermal cyst. The patient was discharged four days after surgery without complication. No tumor recurrence was seen on CT 11 months after the operation (Figure 5.1).

Case 3

A 37-year-old female patient, LYW, was admitted with complaints of difficulty in emptying during bowel movements. On bimanual vaginal and rectal examination, a soft tissue tumor was palpated beside the rectum, with the inferior aspect felt 3 cm above the hymen. The superior aspect of the tumor was not able to be reached. Biochemical markers as mentioned previously were within normal limits with the exception of a slight elevation of CA-153 (25.35U/mL, normal range 0 U/mL to 25 U/mL). Transvaginal Doppler ultrasonography (Figure 1.3) demonstrated a soft tissue tumor measuring 97 mm × 71 mm × 72 mm that was posterior and to the right side of the rectum. The tumor had a smooth wall with scattered hyperechoic content in it and normal blood flow surrounding it. On CT, the apex of the tumor was at the level of the superior articular facet of S4 while the inferior aspect was below the tip of the coccyx (Figure 2.3). The tumor located

to the right of the sacrum without adhesions in the horizontal plane (Figure 3.3).

Similar to Case 2, the tumor could only be seen partially at the time of laparoscopy (Figure 4.3). The superior aspect of the tumor was dissected laparoscopically. However, the inferior aspect of the tumor could not be reached laparoscopically, and the surgery was converted to a transvaginal approach and is further described in the discussion below. Estimated blood loss was 100 mL and duration of the operation was 295 minutes. The final pathological diagnosis of specimen was reported as epidermoid cyst. The patient was discharged home on postoperative day 7. She had no intra- or postoperative complications. No tumor recurrence was seen on CT scan 22 months after the operation (Figure 5.2).

Case 4

A 53-year-old woman, XLM, was admitted due to an asymptomatic pelvic mass. On bimanual vaginal and rectal examination, both the superior and inferior aspect of the tumor could be palpated. Cancer-related biochemical markers described previously were all within the normal range. Transvaginal Doppler ultrasonography (Figure 1.4) demonstrated a soft tissue tumor measuring 65 mm × 48 mm × 43

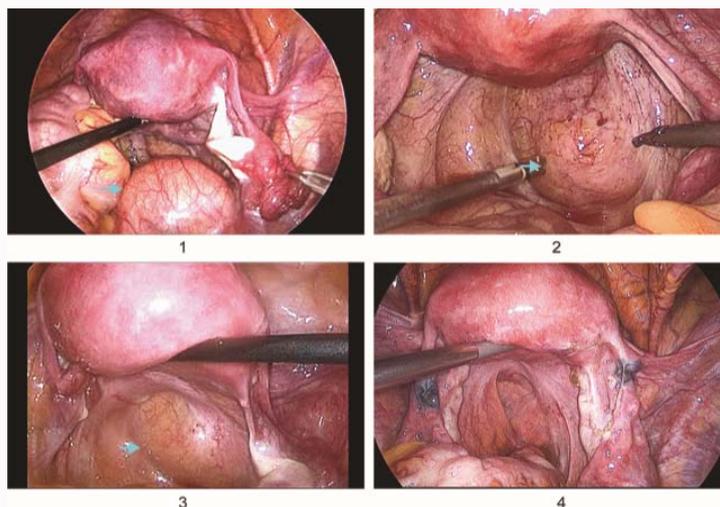


Figure 4: The anatomical findings of benign presacral tumors during the previously described surgeries. The tumors are marked by the blue arrows.

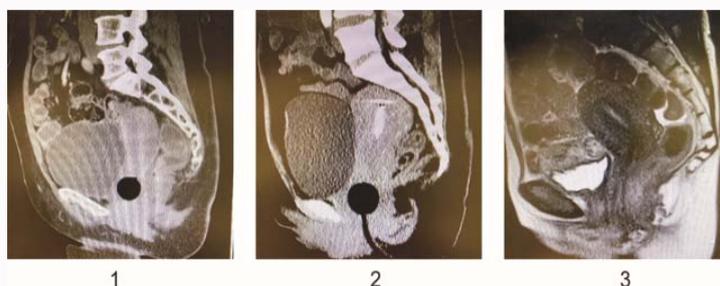


Figure 5: Repeated postoperative imaging results after surgical resection of benign presacral tumors on CT/MRI.

mm, located behind the right aspect of the rectum. It had a smooth wall with scattered hyperechoic content and normal blood flow surrounding it. On MRI (Magnetic Resonance Imaging), the superior aspect of the tumor was at the level of the superior articular facet of S5 while the inferior aspect was about 3 cm below the tip of the coccyx (Figure 2.4). The tumor was located to the right of the sacrum without adhesions in horizontal plane (Figure 3.4).

The tumor could not be seen at the time of laparoscopy (Figure 4.4), and the entire dissection of the tumor was performed through the transvaginal approach. Estimated blood loss was 30mL and the duration of operation was 139 minutes. The final pathological diagnosis of specimen was reported as epidermoid cyst. This patient complained of severe hip pain that interfered with her ability to ambulate postoperatively. However, her pain was relieved two days after having acupuncture treatment and did not recur. She was discharged home on postoperative day 7. No tumor recurrence was seen on MRI 6 months after the operation (Figure 5.3).

Discussion

Strategies to selection of surgical approach

The specific surgical approach should be determined by the position of the tumor, the relationship with adjacent structures, the benign or malignant appearance of the tumor, and its size [12]. It has been proposed that the abdominal approach should be performed for tumors located above the middle of the 3rd or 4th sacrum (S3 or S4) [13,14], while the posterior route should be reserved for those below these levels [15]. For tumors encompassing multiple levels, a

combined approach is recommended [16]. However, unique female anatomy allows for consideration of the transvaginal approach.

Due to emerging technology and further development and refinement of vaginal surgical techniques, we have increasingly recognized that the transvaginal approach may be competitive in terms of consideration for different surgeries in different surgical disciplines [17-19]. We believe that the transvaginal approach has the potential to play a role in the resection of benign presacral tumors.

Here, we propose a decision-making strategy for determination of surgical route in resection of benign presacral tumors in female patients [20]. We describe 5 types of tumors based on their location with considerations for surgical approach in each tumor. In the context of normal female anatomy, the fifth sacrum (S5) is approximately at the same level as the recto-uterine pouch or the posterior fornix.

Type 1: Tumors with the inferior aspect higher than S5 can be completely exposed laparoscopically and tumor removal can be done through laparoscopic surgery.

Type 2: For tumors, whose superior aspect is above S5 and inferior aspect higher than the tip of coccyx, resection can also be done laparoscopically and, if necessary, pushing the tumor transvaginally can provide better exposure to allow for complete resection.

Type 3: For tumors with the superior aspect above S5 and inferior aspect below the tip of coccyx, surgery should be performed through the combined laparoscopic-vaginal approach.

Type 4: For tumors whose superior aspect is below S5, transvaginal

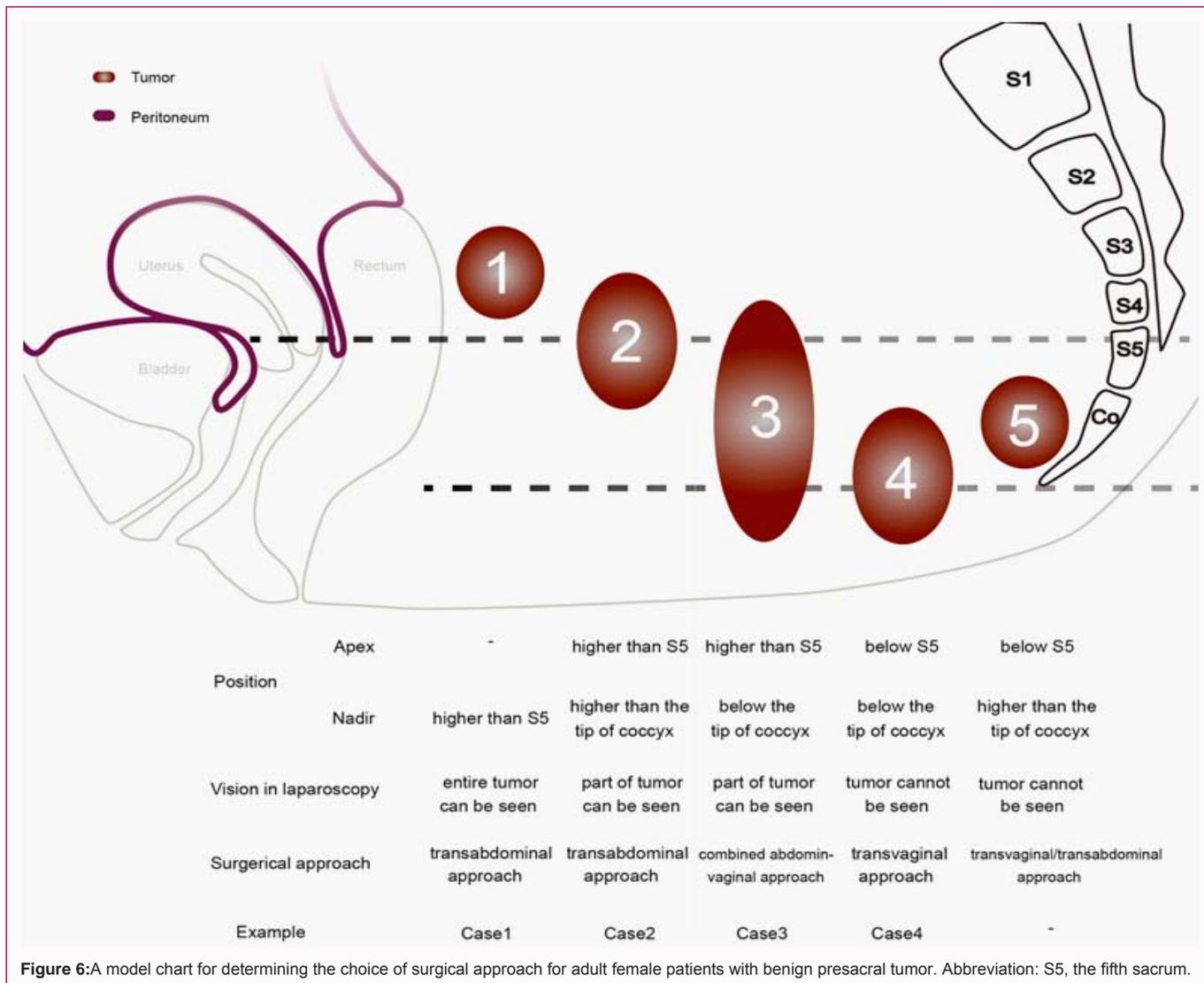


Figure 6:A model chart for determining the choice of surgical approach for adult female patients with benign presacral tumor. Abbreviation: S5, the fifth sacrum.

surgery should be considered.

Type 5: Tumors whose superior aspect is lower than S5 and inferior aspect is higher than the tip of coccyx are rare. They are difficult to be detected because most patients are asymptomatic. Surgical approach should be based on the judgment of the surgeon.

The feasibility and safety of different approaches

The laparoscopic approach: The laparoscopic approach is one of the minimally invasive approaches for resection of presacral tumors, which offers benefits over laparotomy including improved visualization with the laparoscope and smaller abdominal incisions [15].

Over the years, an increasing number of studies have illustrated the feasibility and safety of laparoscopic techniques in the removal of benign presacral tumors. In 1995, Sharpe et al. [21] reported the first laparoscopic removal of a benign pelvic retroperitoneal dermoid cyst with excellent surgical outcome. Eight years later, Köhler et al. [22] successfully completed laparoscopic removal of a large presacral ganglioneurofibroma with no postoperative sensory or motoric dysfunction. A comprehensive literature review [23] of 341 studies demonstrated that presacral tumors in 83 cases were removed through

the laparoscopic approach with a lower recurrence rate than open surgery (0 vs. 21.2%; p<0.05). One explanation for lower recurrence rates in laparoscopic surgery includes fewer laparoscopic cases for malignant tumors compared to laparotomy. A French multicentric study [24] described 270 consecutive patients who underwent presacral tumor removal. 27% of these surgeries were performed through an abdominal approach and was significantly associated with a higher risk of postoperative ileus (7.5%) and rectal fistula (6%). Therefore, despite some acceptable shortcomings, laparoscopy is a widely-used and universally accepted approach for benign presacral tumor removal.

The transvaginal approach: The posterior approach is commonly used in presacral benign tumor removal. Colorectal or orthopedic surgeons who perform most of these cases may not be as familiar with the anatomy of the transvaginal approach compared to gynecologists, and that may be one of the reasons why the transvaginal approach is not as widely performed [10].

However, the transvaginal route has been demonstrated as feasible and safe for a variety of surgeries [17,25]. It has been reported that a transvaginal route can provide better maneuverability and is suitable in various surgeries involving upper-abdominal organs (e.g. the liver,

Table 1: The description of symptoms, signs, auxiliary examinations, operation situation of 4 patients involved.

Case		1	2	3	4
Name		JZL	HYM	LYW	XLM
Age (year-old)		43	49	37	53
Symptom		none	none	Tapered stool	none
Physical examination	Apex Nadir	Cannot be reached	Cannot be reached 4 cm above the hymen	Cannot be reached 3 cm above the hymen	Can be reached 3 cm above the hymen
Serum cancer biomarkers*		normal	normal	CA153 25.36 U/ml (normal interval: 0-25)	normal
Doppler ultrasound	boundary	clear	clear	clear	clear
	size	50 mm × 49 mm × 40 mm	51 mm × 44 mm × 47 mm	97 mm × 71 mm × 72 mm	65 mm × 48 mm × 432 mm
	wall	smooth	smooth	smooth	smooth
	content	mixed structure with scattered increased echo	mixed structure with scattered increased echo	mixed structure with scattered increased echo	mixed structure with scattered increased echo
	CDFI	normal blood flow	normal blood flow	normal blood flow	normal blood flow
CT/MR scan	apex	at the same level as superior articular facet of S1	at the same level as superior articular facet of S4	at the same level as superior articular facet of S4	at the same level as superior articular facet of S5
	nadir	at the same level as superior articular facet of S4	at the same level as the tip of coccyx	below the tip of coccyx	below the tip of coccyx
	relationship with the midline	on the right side of the midline	on the right side of the midline	on the right side of the midline	on the right side of the midline
	Relationship with sacrum	no adhesion	no adhesion	no adhesion	no adhesion
Surgical approach		a laparoscopic surgery	A vaginal-assisted laparoscopic surgery	a combined abdomino- vaginal surgery	a transvaginal surgery
Pathological result		schwannoma	epidermoid cyst	epidermal cyst	epidermoid cyst
Following-up		Lost to follow up	No recurrence was found 22 months after the operation	No recurrence was found 11 months after the operation	No recurrence was found 6 months after the operation

*Serum cancer biomarkers include CEA, AFP, CA153, CA199, CA125, HE4, SCC, NSE
CDFI: Color Doppler Flow Imagination

gallbladder, spleen, abdominal esophagus, or stomach) [26] and even retroperitoneal organs (e.g. the kidney) [27,28]. Additionally, it takes advantage of a natural orifice and can minimize the functional and esthetic impact compared to other cutaneous accesses [6].

Applying transvaginal surgery to removal of presacral tumors has been reported in 2 case reports [10,11]. In this study, we had two similar cases as described above. Among all of our 4 cases, complete resection of each presacral tumor was successful, with no recurrence seen after follow up. The only complication described was acute postoperative hip pain, which has also been seen with Kraske modified procedures for presacral tumors [24] and may result from injury of pelvic plexuses during surgery. Based on these cases, we believe that the transvaginal approach is a safe and feasible method for removal of benign presacral tumors; however, long-term studies with a larger population are needed (Table 1).

We also discuss some tips and tricks from our limited cases. Firstly, the entire dissection of the tumor must be done as close as possible to the tumor capsule, and it should be removed as completely as possible. Secondly, if the tumor is relatively big, the position of the ipsilateral ureter and internal iliac vessels should be identified or exposed in order to avoid inadvertent injury. Thirdly, in the process of dissecting the medial side of the tumor, the surgeon can put the index finger into the rectum in order to discern the relationship between tumor and rectum which may prevent the rectum, the mesorectum and the lateral rectal ligament from being damaged. Fourthly, when dissecting the posterior aspect of the tumor, great attention should be paid to the location of the presacral fascia. The presacral fascia is the thickened part of the parietal pelvic fascia, covering the sacrum, coccyx, nerve, median sacral artery and anterior sacral vein. These

veins, having no valves, communicate widely with the intraspinal venous system. Surgical injury to the presacral fascia can cause uncontrollable bleeding of the presacral vein. Although utilization of electrocoagulation, which is an efficient and effective hemostatic method, can be used to maintain a hemostatic operative field, suture ligation may more suitable any bleeding pedicles close to the sacrum. Fifthly, after the tumor is removed completely, the wound should be repaired by purse-string suture using 2-0 absorbable suture, in order to protect adjacent vessels, nerves, or ureters from being injured.

Application of transvaginal natural orifice transluminal endoscopic surgery (vNOTES)

Due to advancement of surgical techniques and instrumentation, transvaginal NOTES surgeries are gaining popularity. Two major advantages of vNOTES surgery compared to traditional laparoscopy and transvaginal surgery are no abdominal incisions with improved cosmesis and improved visualization through a laparoscope. Application of vNOTES can improve performance of gynecologic surgeries [29,30] and may be a viable option for the surgical treatment of various vaginal complications or pathology, such as vaginal mesh erosion [31], vaginal septum [32], upper vaginal leiomyoma [33], and apical prolapsed [34].

The normal retrorectal space is limited. As the presacral tumor grows larger, it can distort normal presacral anatomy and approach the vagina. vNOTES may be a feasible approach for the removal of presacral tumor; however, no reported cases of resection of presacral tumors *via* vNOTES has been described but may be worth exploring in the future.

Limitations

The limitations of this study include: its retrospective nature, low

case number, and description of different approaches to resection without standardization. In addition, we were unable to assess for any follow up long term issues in case 1. However, the laparoscopic approach that this patient underwent is a commonly used approach, and many similar cases have reported in different studies with relatively good long-term prognosis [24,35]. The most notable strengths in our study includes the introduction of the transvaginal approach to resection with success and proposal for decision making guidelines based on tumor location.

Conclusion

This study describes strategies in determining of surgical approach for the resection of benign presacral tumors and also hopes to increase attention to transvaginal surgery in removal of benign presacral tumor of adult women. However, larger case studies are needed to validate this strategy and to determine the best surgical routes. The gynecologist can be an important participant in presacral tumor removal surgery due to their familiarity of female anatomy.

Ethics Approval and Consent to Participate

This study was approved by the Ethics Committee of Guangdong Provincial Hospital of Chinese Medicine. Informed consent to participate in the study was obtained from all participants. The study was conducted in compliance with the study's protocol and in accordance with the provisions of the Declaration of Helsinki (2013).

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IRB Approval

This research has been approved by the IRB review at the Second Affiliated Hospital of Guangzhou University of Chinese Medicine.

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