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# Diagnostic and Surgical Dilemmas of Pararectal Masses - A Case Series of Unusual Presentations of Extrauterine Leiyomyomas with Review of Literature

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**Abstract**

Extrauterine leiomyomas are rare, and reports of their appearance in the rectovaginal and retrorectal spaces are scarce. Masses in these regions impose diagnostic dilemmas and surgical challenges as there are several differential diagnoses of which some may be malignant. Also, these areas are often difficult to biopsy and technically challenging to approach surgically. We share our clinical experience treating two patients with retrorectal and rectovaginal septal masses. Both patients were in their 50s and presented with chronic lower abdominal pain and menorrhagia with incidental findings of retrorectal and rectovaginal masses on physical and radiological examination. Differential diagnoses were gastrointestinal stromal tumor, endometriosis, benign mesenchymal tumor, neurogenic tumor, adenocarcinoma and sarcoma. Both underwent successful surgical excision of the lesion with organ preservation and the final histology confirming extrauterine leiomyoma. We aim to share an overview of the clinical presentation, diagnostic challenges, and management options for rectovaginal and retrorectal masses, focusing on surgical approaches. We also present a comprehensive review of this rare but important clinical topic.

**Keywords:** Extrauterine fibroid; Extrauterine leiomyoma Pararectal mass; Rectovaginal mass Retrorectal mass

**Abbreviations:** TAMIS: Trans Anal Minimally Invasive Surgery; TLHBSO: Total Laparoscopic Hysterectomy and Bilateral Salpingo-Oophorectomy

## Introduction

Rectovaginal and retrorectal masses are relatively uncommon clinical conditions that can present with nonspecific symptoms such as pelvic pain, dyspareunia, constipation and urinary urgency, or may be discovered incidentally on imaging studies performed for other reasons. The differential diagnosis of masses in these regions is broad and includes a range of benign and malignant conditions [1-7]. Depending on the benign or malignant nature of the mass, surgical approaches will differ drastically depending on whether organ preservation or enbloc-oncological resection is required. Acquiring an accurate diagnosis with imaging, pre-operative biopsy, and deciding on the optimal surgical intervention is vital to preservation of the rectum where possible, given its essential role in defecation [8-11]. We aim to provide an overview of the assessment, diagnosis, and treatment of retrorectal and rectovaginal masses by highlighting two unusual cases of extrauterine leiomyomas. We will also discuss the challenges and controversies of managing this rare and complex clinical entity [12].

Extrauterine leiomyomas are rare, and they may occur as single, multiple, or even disseminated leiomyomatosis throughout abdominal cavities [13-16]. They have commonly reported locations in the pelvis, including the omentum, peritoneum, bowels, and ovaries [17-20]. Reports of their appearance in the rectovaginal and retrorectal spaces are scarce [21]. Several reports suggest an association with laparoscopic morcellation of specimens post-myomectomy or hysterectomy. These lesions are called iatrogenic parasitic leiomyoma, first described by Kelly and Colon in 1909. These fibroid tissue detaches from the uterus and attaches to another organ, such as the ovaries, fallopian tubes, or pelvic cavities. The implanted fibroid then grows and receives its blood supply from the new location leading to parasitic growth. It remains a diagnostic challenge, given the unusual locations it may occur in. Fortunately, given its benign nature, excision

or enucleation with organ preservation can be performed successfully with a low incidence of local recurrence.

## Case Presentation

### Case 1

A fifty-two year old Chinese lady with a history of previous laparoscopic myomectomy and morcellation of specimen in 2009 presented with chronic lower abdominal pain and post-menopausal bleeding to the gynecology clinic. Vaginal examination showed a 3cm posterior vaginal smooth mass. A digital rectal exam showed an anteriorly located submucosal mass in the low rectum 4 cm from the anal verge that felt adherent to the rectum. Pelvic ultrasound showed a solid nodule with vascularity measuring 3.4 x 2.8 x 2.7 cm at the right posterior wall of the vagina, as well as a subserosal fibroid measuring 7.3 x 6.9 x 5.7cm at the left anterior wall of the uterus. An MRI of her pelvis showed a 3.4cm perineal mass in the right low rectovaginal septal region. However, the mass was not invading the posterior vaginal wall, and there were no aggressive radiological features.

Colonoscopy was normal confirming that the mass was extraluminal. Differential diagnoses were that of a gastrointestinal stromal tumor, endometriosis, benign mesenchymal tumor, adenocarcinoma, and sarcoma. Given the possibility of both benign and malignant pathologies, and that the extent of surgical resection would depend on the histology, the patient was counseled and underwent examination under anesthesia with a transrectal biopsy of the rectovaginal septal nodule under general anesthesia. Intra-operatively, the mass appeared submucosal on the rectal examination and no mucosal abnormality was identified in the vagina. The mass was 1cm above the anorectal junction and 4cm from the anal verge. An incision was made on the rectal mucosa overlying the mass, and a biopsy was performed using a trucut needle with a satisfactory tissue core obtained for histology. The mucosal defect was sutured closed with Vicryl 2/0 interrupted sutures. Histology showed smooth muscle tumor favoring uterine-type leiomyoma.

Given the benign histology, the patient was planned to undergo total laparoscopic hysterectomy with bilateral salpingo-oophorectomy for her large uterine fibroid, and synchronous excision of the rectovaginal septal nodule. The patient was under general anesthesia in Lloyd-Davies' position. Intra-operatively a large 6cm fibroid was seen on the anterior wall of the uterus. After removing the uterus, we identified two nodules over the rectovaginal septum. A 3.5cm well-encapsulated nodule abutted the low rectum and vagina just above the anorectal junction, while another smaller 1.5cm nodule was found in the right pararectal region attached to the right pelvic floor muscle. The peritoneum over the Pouch of Douglas was incised and the rectum and vagina was separated in the rectovaginal septum down to the pelvic floor. Both lesions were dissected off the rectum and the posterior vaginal wall without rupturing the capsule, then excised sparing the rectum.

The specimens were retrieved using an Endo pouch and extracted through the vagina. The final histology of the resected pararectal nodules showed two smooth muscle tumors. The larger nodule showed a high mitotic count with cellular atypia; however, Ki-67 immunoactivity was low at 5%, and ALK 1 immunostaining was negative. Hence it was labeled as a smooth muscle tumor with uncertain malignant potential. The smaller nodule was a conventional leiomyoma. The patient recovered well after surgery and was discharged on post-operative day 5. Clinical and radiological surveillance showed no sign of recurrence at 47 months of follow-up. A short video of the surgery can be viewed using the following link (Figure 1-3).

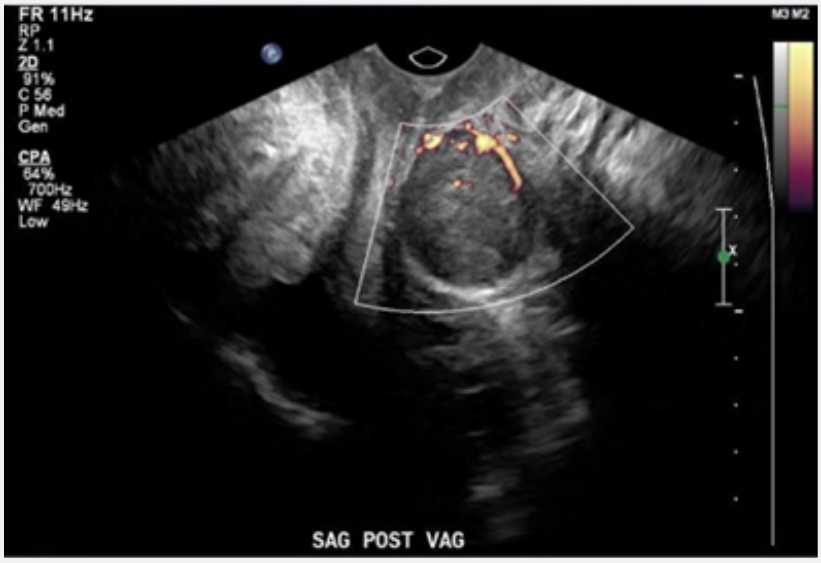


**Figure 1:** Ultrasound image showing posterior vaginal mass with increased vascularity.


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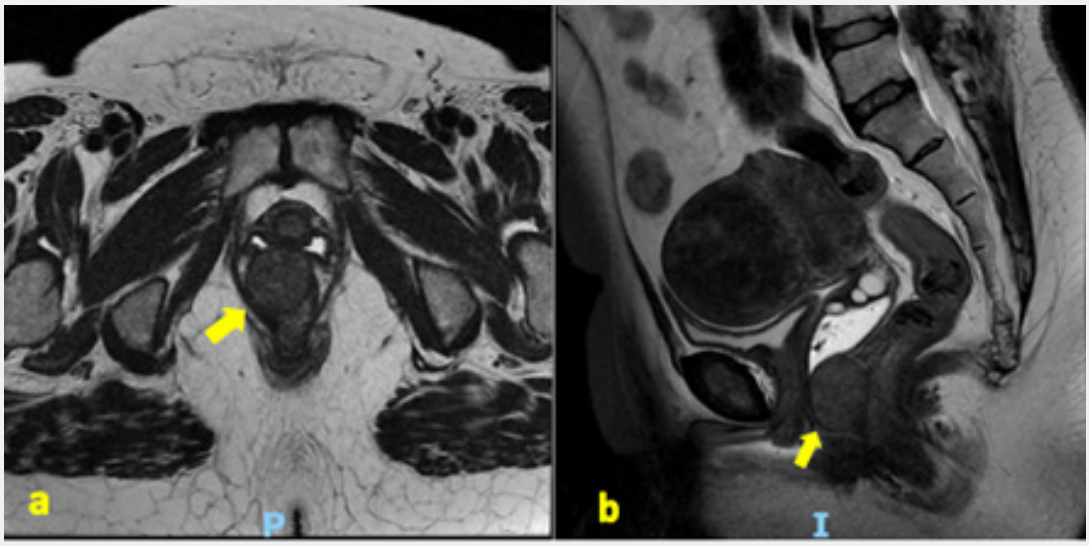


**Figure 2:** MRI pelvis image showing T2 isointense lesion with restricted diffusion (a: axial; b: sagittal).

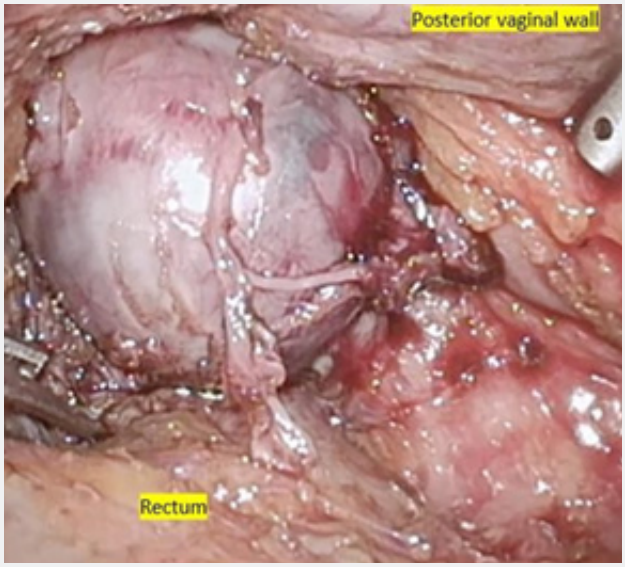


**Figure 3:** Intraoperative appearance of 3.5cm rectovaginal mass.

## Case 2

A fifty-one year old Chinese lady with a history of previous laparoscopic myomectomy and morcellation of specimen in 2003 presented with chronic lower abdominal pain and perimenopausal menorrhagia at a gynecology clinic. Digital rectal examination revealed a 5cm left poster-lateral pararectal mass with its lower limit at 6cm from the anal verge. It was well circumscribed, immobile and not invading the rectal lumen. Ultrasound of the pelvis showed a well-defined, heterogenous mass measuring 4.4 x 3.9 x 3.1cm with prominent arterial and venous vascularity. There were also multiple uterine fibroids seen ranging between 3-4cm. MRI of the pelvis showed a poster-lateral retrorectal solid mass anterior to the sacrum but separate from the rectum and cervix, measuring 5.0 x 3.8 x 3.0cm. Colonoscopy was normal. Differential diagnoses were neurofibroma, schwannoma, benign mesenchymal tumor, and sarcoma. Biopsy was not done in this case as the tumor was separate from the rectum and difficult to access via percutaneous or transrectal routes. Although its radiological features appeared benign, with a clear tissue plane between the rectum and the mass, retrorectal masses ares associated with a significant risk of malignancy.


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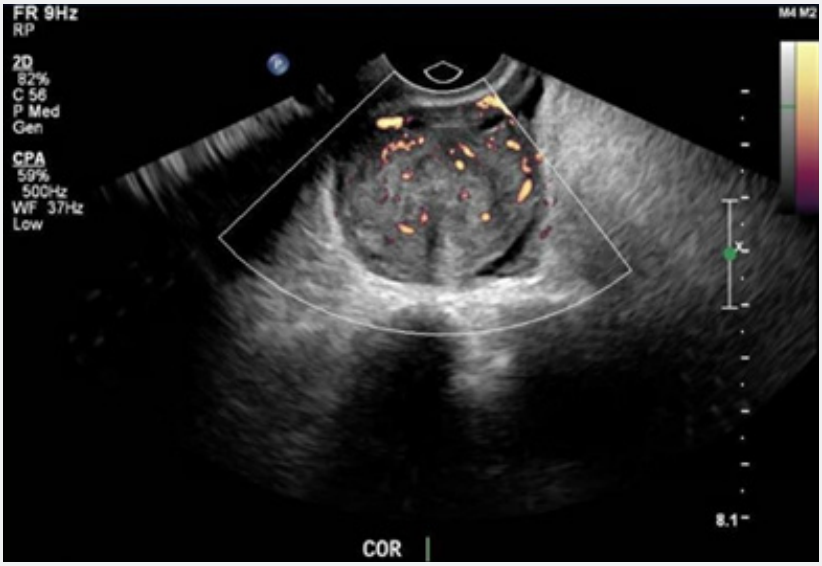


**Figure 4: Ultrasound image showing well-defined but heterogenous mass with prominent vascularity.**

Hence, the patient was offered surgical excision of the lesion The patient underwent total laparoscopic hysterectomy and bilateral salpingo-oophorectomy (TLHBSO), with concurrent excision of the retrorectal mass. Intra- operatively, the patient had a bulky 16 week sized uterus with multiple fibroids. The retrorectal mass was well-circumscribed, and although adherent to the mesorectum and levator ani muscles, it was free from the rectal wall. The mass was approached laparoscopically via an incision over the peritoneum of the left pararectal space, dissected free from the mesorectum and pelvic floor muscles, and retrieved together with the resected uterus using an Endo pouch extracted through the vagina. Postoperative histology showed benign leiomyoma. The patient recovered well after surgery and was discharged post-operative on day 5. Clinical and radiological surveillance showed no sign of recurrence at 25 months of follow-up (Figure 4 & 5).

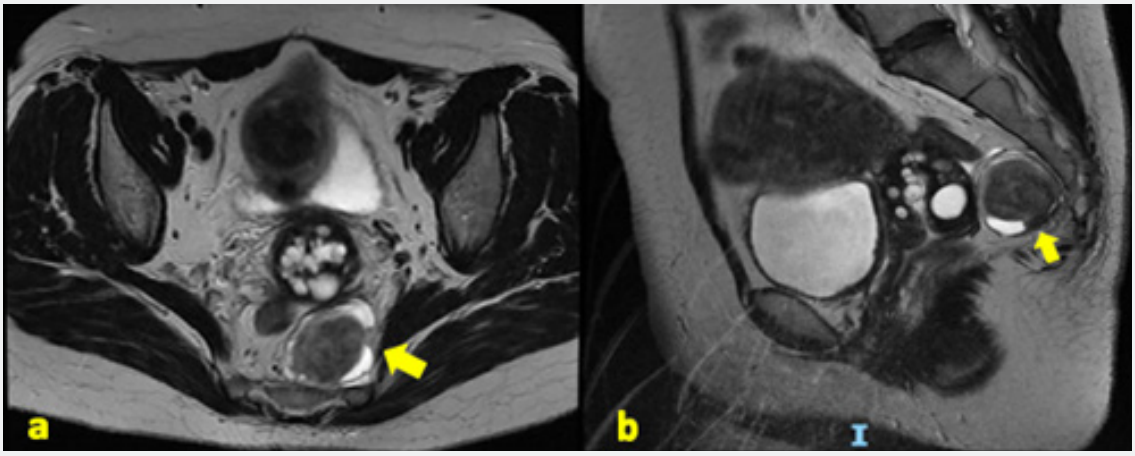


**Figure 5:** MRI image showing T2 isointense mass with central soft tissue component and cystic clefts (a: axial; b: sagittal).

## Discussion

The incidence of retrorectal and rectovaginal septal masses is low, with only case reports and a few case series available in the literature. No guideline or treatment algorithm is available to guide the management of this clinically challenging entity. Not to mention that the spectrum of diseases that arises from this anatomical region can be wide-ranging, from gastrointestinal


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stromal tumor, endometriosis, benign mesenchymal tumor, neurogenic tumors, to adenocarcinoma and sarcoma. Furthermore, their non-specific radiological findings often require histological correlation to determine pathology. In both cases, patients had a significant past surgical history of laparoscopic myomectomy and morcellation of specimens. Although many have reported an association between laparoscopic morcellation with parasitic leiomyoma, reports of extrauterine fibroids in the rectovaginal septum and retrorectal space are rare. These two anatomical spaces are not intra-peritoneal and are not dissected during the myomectomy procedure; hence less likely for parasitic seeding. The other reported case by AlShalabi O et al. [21] did not have a history of myomectomy or morcellation. Some authors have proposed two other possibilities, including tumorigenesis occurring de novo from cells within the vascular wall or through the invasion of a uterine leiomyoma into the vessel itself [22-23]. Benign metastasizing leiomyoma has also been theorized in some cases, with reports of having existing uterine leiomyoma or previous surgical procedures related to uterine leiomyoma.

However, the most commonly reported location was the lung, with other sites including the heart, bones, liver, lymph nodes, bladder, skeletal muscles, and central nervous system [24-28]. Nevertheless, these phenomena of extrauterine leiomyoma remain a challenge for experts. There are several approaches reported for tumors in the rectovaginal septum. I. Anev et al. [4] described the open transvaginal approach via an incision over the posterior vaginal wall directly over the mass, followed by primary defect repair.4 AlShalabi O et al. [21] described a combined open transabdominal and transperineally approach for a large 17cm mass enucleation [21]. Trans anal approaches including trans anal minimally invasive surgery (TAMIS) can also be considered if the tumor is located in the low rectum. However, this will create a full-thickness rectal defect which needs to be repaired and bears a risk of wound breakdown and pelvic sepsis. Given that our first patient also required a hysterectomy for her large uterine fibroid, we chose the laparoscopic transabdominal approach as the tumor size was favorable and allowed synchronous resection of both pathologies. The technical challenge was dissecting laparoscopically in a narrow space in the low pelvis to excise the rectovaginal septal nodules while not injuring the rectum or vagina. In the first patient, pre-operative histology confirmed a benign pathology, hence there was no necessity to excise the previous transrectal biopsy tract. As the disease was benign, we were able to extract both the uterus and rectovaginal masses using natural orifice extraction via the vagina without the risk of tumor seeding, hence optimizing post operative recovery.

Retrorectal masses, have conventionally been described to be removed using the Krake procedure, a transperineally approach with the patient in a prone-jackknife position. A vertical incision is made from the lower sacrum to the upper border of the anus, with the anococcygeal ligament divided and the levator ani muscle retracted laterally to gain access to pre-sacral space [29]. A sizeable single-center cohort study with 143 patients over thirty years, from 1990 to 2019, has reported 14 malignant cases out of 107 operated patients [30]. This study has shown that a trend toward a laparoscopic approach likely coincides with the widespread adoption of laparoscopy in colorectal surgery. Similar to our previous case, a transabdominal minimal invasive approach was chosen in our second patient to ease synchronous resection of the uterus and excision of the retrorectal lesion. Other approaches that can be considered include a transrectal open or minimally invasive approach depending on the proximity of the mass to the rectum. However if this approach were to be adopted, primary closure of the rectal defect will have to be performed. Also, if a pre-operative biopsy was performed, consideration needs to be taken of excision of the biopsy tract during the surgery. We propose a simple algorithm below for the management of pararectal masses, which we hope will be useful to surgeons encountering this rare entity.

## Conclusion

Extrauterine leiomyomas remain rare, with scare reports of their appearance in the rectovaginal septum and retrorectal space. These cases are best managed with multidisciplinary input in a specialist center with advanced laparoscopic capability. Management of these cases will require comprehensive assessment using ultrasound and MRI imaging with or without biopsy, depending on the clinical diagnosis and feasibility. Modern surgical resection techniques include transabdominal and trans anal approaches which can be performed laparoscopically. We conclude that in our experience, these lesions can be safely excised laparoscopically sparing the rectum, with a low risk of local recurrence.

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