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Case analysis and literature review of rectal wall dissemination following laparoscopic myomectomy

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Abstract. Uterine leiomyomas are the most common benign gynecologic tumors of the female reproductive system. Laparoscopic minimally invasive surgery has become a critical surgical procedure for symptomatic uterine leiomyomas. With the widespread use of uterine morcellators during laparoscopic myomectomy, the rare late complication associated with this technique, Leiomyomatosis Peritonealis Disseminata (LPD), has garnered increasing attention among gynecologists. This paper reports one case of LPD admitted to the Department of Gynecology of Zhuhai People's Hospital Affiliated to Jinan University (hereinafter referred to as our hospital) and reviews the existing literature. LPD is a potential devastating consequence of unrestricted laparoscopic morcellation. For the surgical treatment and management of LPD, especially in cases involving multiple organ involvement, a multidisciplinary diagnosis and treatment approach is clinically recommended, with surgery as the first-line treatment.

Keywords: leiomyomatosis peritonealis disseminata, morcellator, iatrogenic, laparoscopic myomectomy

1. Introduction

Uterine leiomyomas are the most common solid benign gynecologic tumors of the female reproductive system, detected in 70%–80% of women during their lifetime. Up to 70% of women with uterine leiomyomas are asymptomatic, while approximately 30% of patients develop severe complications [1]. Uterine leiomyomas are associated with infertility and other adverse obstetric outcomes. For symptomatic patients desiring fertility preservation, laparoscopic myomectomy serves as a valuable therapeutic strategy. Characterized by mild postoperative pain, rapid recovery, cosmetic outcomes and favorable reproductive effects, laparoscopic surgery is recognized as the mainstream surgical approach in this field [2]. However, large uterine leiomyomas require morcellation for removal from the abdominal cavity. Some fragments or small leiomyomas may be inadvertently left in the abdominal cavity during removal, subsequently implanting throughout the peritoneum, deriving blood supply from adjacent tissues, and growing into parasitic leiomyomas with diverse clinical manifestations [3]. This paper reports a case of recurrent leiomyoma on the rectal serosa following laparoscopic myomectomy in a 31-year-old female patient and briefly reviews the

current relevant literature. It aims to explore the clinical characteristics, prevention and treatment of iatrogenic LPD, so as to enhance the understanding of LPD and facilitate its clinical management.

2. Case report

2.1. History and physical examination

A 31-year-old female patient was admitted to the Department of Gynecology of our hospital on October 14, 2023, due to a gradually enlarging uterine mass detected on physical examination for over one year. In 2020, she underwent laparoscopic myomectomy and left mesosalpinx cyst resection at the Fifth Affiliated Hospital of Zunyi Medical University for uterine leiomyomas. Intraoperatively, a protruding mass measuring approximately 6 cm × 5 cm × 6 cm was observed on the upper-middle anterior uterine wall, with an irregular surface, engorged blood vessels, and a pedicle about 5 cm thick extending into the myometrium. A 2 cm × 2 cm cyst was found on the left mesosalpinx. After complete enucleation of the leiomyoma, rotational morcellation of the leiomyoma was performed and the tissue was extracted through the abdominal wall trocar site without placing the specimen in a laparoscopic specimen bag. Intraoperative frozen-section pathology confirmed uterine leiomyoma with ischemic necrosis. In 2022, gynecological ultrasonography at the Fifth Affiliated Hospital of Zunyi Medical University indicated a uterine leiomyoma of approximately 3 cm, which was non-palpable on gynecological examination. The patient presented with no menstrual changes, abdominal pain or distension, and was advised to undergo regular follow-up without special intervention. In March 2023, repeat ultrasonography at the same hospital showed an enlarged uterine leiomyoma of 6–7 cm. The patient presented to our hospital on August 28, 2023, and pelvic gynecological ultrasonography (Figure 1) revealed a hypoechoic mass in the uterine myometrium, located on the posterior wall (Type VII), measuring approximately 104 mm × 70 mm × 78 mm, roughly round with a relatively clear boundary. The imaging diagnosis suggested a solid uterine lesion, suspicious for uterine leiomyoma. For further treatment, the patient was admitted with a provisional diagnosis of "pelvic mass" on October 14, 2023. The patient had been in good general health, with no history of hypertension, diabetes or coronary atherosclerotic heart disease. She underwent laparoscopic myomectomy at the Fifth Affiliated Hospital of Zunyi Medical University in April 2020 and segmental resection and biopsy of both breasts on August 30, 2023, with postoperative pathology confirming breast fibroadenoma. No history of blood transfusion. Other medical history included intermittent anxiety, for which she received regular outpatient medication at the Psychology Department of our hospital. She discontinued the medication six months prior to admission and was emotionally stable at presentation. Born in Guangdong Province, she had no history of blood transfusion, drug allergy, smoking or alcohol consumption, and no history of unprotected sexual contact. No family history of hereditary diseases across two or three generations. Her parents were both alive, with no family history of similar diseases, hemophilia or thalassemia.

2.2. Physical examination

On admission, vital signs were stable. No obvious abnormalities were noted on cardiac, pulmonary or abdominal examination. Gynecological examination revealed a married, nulliparous vulva, patent vagina with no abnormal discharge, smooth, normal-sized, firm cervix with no tenderness on motion. The uterus was enlarged to the size of a 2+ month gestation, with an irregular surface and no tenderness. No abnormalities or tenderness were palpated in the adnexal regions bilaterally.

2.3. Laboratory and auxiliary examinations

Admission blood tests showed hemoglobin 121 g/L, white blood cell count 7.33×10^9/L, platelet count 280×10^9/L. Coagulation function and biochemical tests were within normal limits. Tumor markers: CA125 22.10 U/mL, CA19-9 22.82 U/mL, CEA 0.76 ng/mL, HE4 41.50 pmol/L, SCC 1.20 ng/mL, AMH 5.31 ng/mL. Sex hormone levels: estradiol 879.47 pmol/L, progesterone 36.94 nmol/L, follicle-stimulating hormone 3.78 IU/L. Pelvic gynecological ultrasonography: The uterus was anteverted with abnormal morphology, measuring approximately 66 mm × 43 mm × 40 mm. The endometrium was midline, 9 mm thick with homogeneous echogenicity. Myometrial echogenicity was heterogeneous, with a hypoechoic mass in the posterior wall (Type VII), measuring 104 mm × 70 mm × 78 mm, roughly round with a relatively clear boundary and uneven internal hypoechogenicity. Both ovaries were visualized, with no obvious abnormal echogenicity in the bilateral adnexal regions. CDFI: Punctate blood flow signals were observed in the uterine myometrium; punctate and linear blood flow signals were noted around and within the hypoechoic mass. Contrast-enhanced abdominal Computed Tomography (CT) (Figure 2): An intra-abdominal mass contiguous with the uterus was identified, with a maximum cross-sectional size of approximately 103 mm × 78 mm × 87 mm. Contrast enhancement showed annular and patchy enhancement, with uterine compression and displacement. The uterine space-occupying lesion was highly suspicious for leiomyoma. Gastroenteroscopy (Figure 3): A flat elevated lesion measuring approximately 45 mm × 40 mm was observed at the rectosigmoid junction, about 13 cm from the anal verge, with an ill-defined boundary and surface mucosa consistent with adjacent tissue. The lesion was fixed and pliable on probe palpation. The diagnosis suggested an elevated lesion at the rectosigmoid junction, suspicious for external compression. Chest contrast-enhanced CT, hepatobiliary-pancreatic-spleen ultrasonography, bilateral lower extremity vascular ultrasonography, urinary tract ultrasonography and echocardiography revealed no obvious abnormalities.

2.4. Diagnosis and treatment course

With a provisional diagnosis of pelvic mass of unknown nature, the patient underwent surgical exploration on October 16, 2023. Intraoperative exploration (Figure 4) showed a normal-sized uterus, with a round mass measuring approximately 10 cm × 8 cm × 8 cm on the wall of the rectosigmoid junction. The mass had a smooth surface, firm texture and good mobility. Given the history of previous uterine leiomyoma surgery, a rectal wall leiomyoma was suspected. The entire mass was completely enucleated using an ultrasonic scalpel close to the lesion surface, hemostasis was achieved, and no intestinal perforation was noted. The peritoneum and intestinal serosa were closed with continuous absorbable surgical sutures. The specimen was placed in a laparoscopic specimen bag, extracted via a 5 cm periumbilical incision, and morcellated within the bag for complete removal. Gross examination of the rectal specimen: A pile of irregular muscular tissue measuring 15 cm × 14 cm × 4 cm, with serosa locally visible on the surface, gray-white and smooth. The cut surface was gray-white to gray-red, solid and firm. Routine pathological examination was performed. Postoperative pathological diagnosis (Figure 5): Rectal mass resection specimen: Spindle cell tumor, consistent with leiomyoma with hemorrhage, degeneration, infarction and hyalinization, based on immunohistochemical staining results and histomorphology. Immunohistochemical results (block A9): Vimentin (+), SMA (diffuse +), Desmin (diffuse +), ER (partial +), PR (+), FH (+), 2SC (-), CD34 (-), DOG-1 (-), S-100 (-), Ki-67 (5%, +), CD117 (-), SDHB (+), STAT6 (-), ALK (-), BRAF V600E (-). Special staining results (block A7): Masson (+, collagen deposition observed), reticular fiber staining (-).

2.5. Outcome

The patient had a smooth postoperative course with normal body temperature. Prophylactic antibiotics (ceftriaxone plus metronidazole), acid suppression, analgesia and nutritional support were administered. No endocrine therapy was given, and the patient was discharged on postoperative day 7. At the time of manuscript preparation, the patient had been followed up closely for 2 years postoperatively, remained asymptomatic, and pelvic color Doppler ultrasonography showed no specific abnormalities.

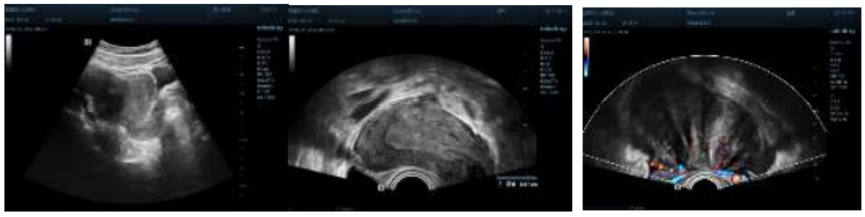

Figure 1. Results of pelvic gynecological ultrasonography

Note: A hypoechoic mass measuring approximately 104 mm × 70 mm × 78 mm was observed in the pelvic cavity, with a roughly round shape, relatively clear boundary, and uneven internal hypoechogenicity.

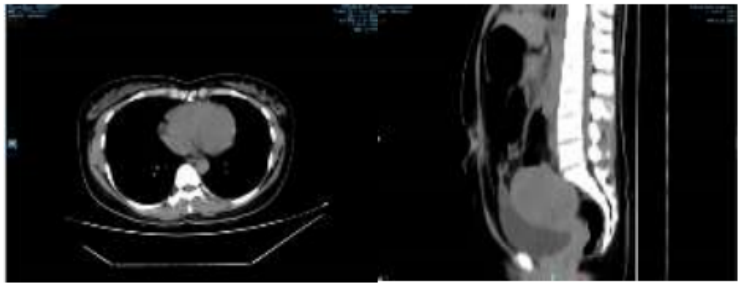

Figure 2. Contrast-enhanced abdominal computed tomography

Note: An intra-abdominal mass contiguous with the uterus was identified, with a maximum cross-sectional size of approximately 103 mm × 78 mm × 87 mm. Contrast enhancement showed annular and patchy enhancement, with uterine compression and displacement.

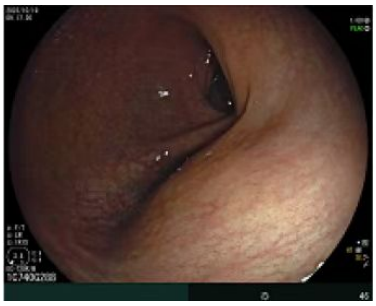

Figure 3. Results of gastroenteroscopy

Note: A flat elevated lesion was observed at the rectosigmoid junction, about 13 cm from the anal verge.

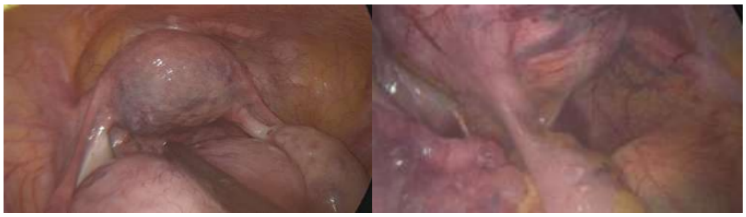

Figure 4. Laparoscopic pelvic exploration

Note: Intraoperatively, the uterus was normal-sized, with a 12 cm × 10 cm × 10 cm mass at the rectosigmoid junction closely adherent to the intestinal wall. Abundant blood supply was noted on the mass surface.

3. Discussion

3.1. Pathogenesis

LPD was first reported and described by Willson and Peale in 1952 [4]. A rare multifocal proliferative disorder, LPD is characterized by multiple leiomyomatous nodules of varying sizes in the peritoneal cavity, commonly involving the uterus, fallopian tubes, intestines, mesentery, omentum and retroperitoneum [4]. LPD predominantly affects women of reproductive age, with only a few cases reported in postmenopausal women. To date, fewer than 200 cases have been documented in the literature [5]. Many cases are asymptomatic, leading to an underestimated incidence due to the lack of systematic follow-up [5].

The pathogenesis of LPD remains unclear, with four potential etiological factors proposed: hormonal factors, metaplastic transformation of subperitoneal mesenchymal stem cells, hereditary factors and iatrogenic factors [6]. High levels of female gonadal steroid hormones play a critical role in the pathogenesis of LPD. Most reported cases involve elevated estrogen and progesterone levels, with a history of pregnancy, long-term oral contraceptive use, or estrogen-secreting tumors [7]. Exposure to high estrogen levels is another risk factor associated with LPD [7]. Spontaneous regression of LPD may occur following the cessation of endogenous or exogenous hormone exposure. Immunohistochemical analysis shows strong expression of estrogen and progesterone receptors in LPD tumor cells, supporting the hypothesis that high estrogen and progesterone levels contribute to LPD pathogenesis [8].

Iatrogenic factors are another important cause of LPD, which may develop following laparoscopic uterine morcellation. During intraoperative rotational morcellation, residual leiomyoma fragments implant on intraperitoneal surfaces, form new blood vessels and establish blood supply with adjacent tissues, leading to the development of iatrogenic disseminated leiomyomas [9, 10]. Previous reports indicate that LPD develops 39–132 months after primary laparoscopic myomectomy, with 1–16 lesions [11]. In recent years, minimally invasive procedures such as High-Intensity Focused Ultrasound (HIFU) ablation and Uterine Artery Embolization (UAE) have emerged as alternative options for patients intolerant to or unwilling to undergo myomectomy. As a non-invasive technique, HIFU safely and precisely ablates uterine leiomyomas without damaging adjacent structures, though no evidence links HIFU ablation to LPD [12]. Batton et al. [13] reported one case of LPD occurring three years after UAE in a patient with no other relevant medical history, warranting further investigation into the causal relationship between LPD and UAE. Additionally, one case of familial clustering of LPD has been documented [14]. In this case, the patient presented with rapidly growing LPD nodules measuring 104 mm on follow-up ultrasonography three years after laparoscopic myomectomy. With no history of estrogen exposure or high expression of estrogen/progesterone receptors in the leiomyoma

nodules, hormonal etiology was considered unlikely. No family history of LPD was reported, confirming an iatrogenic etiology.

3.2. Clinical characteristics

Li J et al. [15] retrospectively analyzed the medical records of 13 LPD patients, with a mean age of 42.23 years (range: 26–51 years). Most patients were asymptomatic, with lesions incidentally detected on physical examination or during surgery. Symptomatic patients presented with non-specific symptoms including abdominal pain, pelvic pain, gastrointestinal symptoms, vaginal bleeding and prolonged menstrual periods [3]. Consistent with literature reports, this patient presented with a progressively enlarging abdominal mass, without pelvic/abdominal pain or abnormal uterine bleeding.

No specific serological markers for LPD have been identified to date. Imaging modalities such as ultrasonography, CT and Magnetic Resonance Imaging (MRI) aid in preoperative diagnosis of LPD, though imaging alone has limited diagnostic value. A detailed medical history is critical for diagnosis. A history of uterine leiomyomas or previous leiomyoma surgery, particularly laparoscopic uterine morcellation, combined with typical imaging features, facilitates accurate preoperative diagnosis.

Diagnosis and differential diagnosis of LPD are challenging due to the absence of specific clinical manifestations (or asymptomatic status) and atypical imaging features, leading to a high preoperative misdiagnosis rate. LPD is frequently misdiagnosed as multiple leiomyomas or metastatic malignant pelvic tumors. Definitive diagnosis requires intraoperative exploration, postoperative standard histopathological examination and immunohistochemical staining, which distinguish LPD from peritoneal metastatic malignancies (e.g., peritoneal carcinoma, metastatic leiomyosarcoma) and benign conditions (e.g., benign metastatic leiomyoma). Grossly, LPD nodules range from 0.5 cm to 25 cm in diameter (typically 2.0–3.0 cm), mostly round, firm and well-circumscribed, with a white to gray-white whorled cut surface and no obvious hemorrhage or necrosis [16]. Microscopically, LPD nodules consist of smooth muscle cells arranged similarly to leiomyomas, with minimal cellular atypia and low mitotic activity [15]. HE staining reveals abundant blood supply in LPD nodules. Immunohistochemical staining shows strong expression of smooth muscle markers (smooth muscle actin [SMA], desmin), confirming similar molecular cytogenetic features between LPD and uterine leiomyomas. Estrogen and progesterone receptors are variably expressed. Negative staining for cytokeratin, S-100 protein, CD10, CD117 and CD34 (markers associated with malignancy or metastasis) aids in differentiation from malignant tumors [8]. The gross appearance, postoperative paraffin pathology and immunohistochemical staining results of this patient's specimen were consistent with literature findings.

Bu Hualei et al. [8] performed immunohistochemical analysis on 10 uterine leiomyomas, 4 LPD cases and 10 leiomyosarcomas, demonstrating distinct phenotypic differences between LPD and uterine leiomyomas. Compared with the general population, LPD showed at least 4-fold amplification of CDK4, MYC, NBN and DAXX copy numbers. In contrast, these four markers were weakly positive or negative in uterine leiomyomas and strongly positive in leiomyosarcomas. The expression profile of LPD was more similar to leiomyosarcoma, suggesting LPD represents an intermediate entity between benign uterine leiomyomas and malignant leiomyosarcomas. Although LPD has a favorable prognosis, it exhibits malignant-like features including recurrence and widespread pelvic/peritoneal dissemination. Previous cases have reported elevated serum CA19-9 and CA125 levels, markers specific for gastrointestinal and ovarian malignancies, leading to misdiagnosis as metastatic pelvic/abdominal cancer [17]. Malignant transformation is a rare complication of LPD, which is easily misdiagnosed as metastasis from an unknown primary tumor in patients without a typical medical history, resulting in false-positive diagnoses [18]. Therefore, close attention should be paid to the

potential malignant risk of LPD during treatment and follow-up, with complete resection of all visible lesions intraoperatively to prevent malignant transformation of residual lesions.

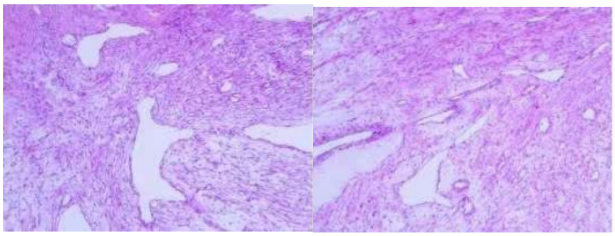

Figure 5. Light microscopic appearance of the resected rectal mass specimen

Note: Rectal mass resection specimen: Spindle cell tumor, consistent with leiomyoma with hemorrhage, degeneration, infarction and hyalinization, based on immunohistochemical staining results and histomorphology.

3.3. Treatment and prevention

There is no international consensus on the treatment of LPD. While surgical resection remains the primary therapeutic approach, decisions regarding uterine and adnexal preservation require individualized treatment plans based on the patient's age and fertility requirements. Given the predilection for women under 40 years of age [5], conservative management with regular systematic follow-up may be considered for small, asymptomatic lesions with no evidence of enlargement.

Pharmacological therapy may include gonadotropin-releasing hormone agonists, aromatase inhibitors and estrogen receptor antagonists to regulate estrogen levels.

For surgical intervention, preoperative evaluation to rule out uterine malignancy is mandatory prior to laparoscopic uterine morcellation, with repeat intraoperative assessment. Open laparotomy and avoidance of morcellation are indicated if malignancy is suspected. Morcellation should be performed within a closed morcellation bag to prevent tissue spillage. Intraoperative care should be taken to avoid injury to intraperitoneal organs, followed by thorough irrigation of the pelvic and abdominal cavities with large volumes of distilled water or normal saline to prevent residual tissue fragments. Some studies recommend limited intraperitoneal irrigation to reduce the spread of tissue fragments to the upper abdomen [19]. These measures do not completely eliminate the risk of iatrogenic LPD, emphasizing the need for enhanced postoperative follow-up protocols with increased surveillance frequency to facilitate early detection and individualized intervention.

While the definitive prevention of LPD remains unclear, the following measures may reduce risk: (1) regular physical examination and screening; (2) healthy lifestyle and dietary habits; (3) maintenance of normal body weight; (4) balanced estrogen levels; (5) avoidance of excessive hormone medication. These recommendations are not definitive preventive strategies but aim to promote overall health and reduce risk factors, with individualized prevention plans tailored to the patient's health status and physician advice.

4. Discussion

Residual uterine leiomyoma fragments following laparoscopic myomectomy can survive and grow at extrauterine intraperitoneal sites under steroid hormone exposure, despite losing uterine blood supply. Most reported cases are asymptomatic, with lesions presenting as abdominal or pelvic masses, leading to an underestimated incidence of iatrogenic parasitic leiomyomas due to inadequate follow-up. There is no consensus on treatment, with both hormonal and surgical therapy effective. Surgical resection is indicated for

large symptomatic lesions. Intraoperative precautions include minimizing excessive tissue fragmentation and performing morcellation within a closed specimen bag whenever feasible. In-bag morcellation may reduce the incidence of iatrogenic disease. The peritoneal cavity should be thoroughly inspected during morcellator use to ensure bag integrity and prevent residual tissue fragments from implanting and forming parasitic leiomyomas. Additionally, patients should be informed of this rare iatrogenic complication and advised to undergo regular clinical follow-up.

5. Conclusion

Although benign, LPD exhibits malignant-like dissemination features and requires comprehensive diagnosis based on clinical history, imaging and histopathology. Surgical resection is the primary treatment, with adjuvant endocrine therapy indicated for select patients and regular clinical follow-up recommended.

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