Case Report

Radical surgery for pseudo-ovarian malignancy: a case report of bowel endometriosis with massive ascites

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Background: Endometriosis is a disturbing condition affecting about 10% of all reproductive aged women. The most severe form of endometriosis is deeply infiltrative endometriosis (DIE). Bowel is commonly affected in DIE. Treatment options of bowel endometriosis include surgery and medication, depending on many factors such as age, the severity of symptoms and desire for pregnancy. At present, the individualized comprehensive management of bowel endometriosis is still under exploration. Here we report an uncommon case of bowel endometriosis treated by radical surgery and postoperative high-dose progestin to enrich the clinical experience.

Case Description: A 37-year-old woman was admitted to our hospital for suspected ovarian malignancy in the presence of pelvic mass, massive ascites and elevated CA-125. A laparoscopic radical surgery was performed, and she was diagnosed with bowel endometriosis. Considering the patient's high recurrence risk indicated by bowel endometriosis, massive ascites, severe adhesions, and dysmenorrhea, six-course gonadotropin-releasing hormone agonists therapy followed by high-dose progestin (two levonorgestrel intrauterine systems and subdermal implants) was administrated postoperatively to improve symptoms and prevent recurrence. No recurrence in bowels was observed by November 2021 (53 months).

Conclusions: Both patient's desire and condition should be considered in the management of symptomatic bowel endometriosis. Optimal surgical removal is of great significance and individualized hormonal therapy may provide an additional component.

Keywords: Bowel endometriosis; ascites; radical surgery; hormonal therapy; case report

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Introduction

Endometriosis is a chronic, recurrent, inflammatory disease defined as the presence of endometriotic glands and stromal cells outside the uterine cavity, disturbing about 10% women of reproductive age (1). It is associated with menstrual irregularities, chronic pelvic pain, dysmenorrhea, and infertility. Endometriomas, peritoneal endometriosis and deeply infiltrating endometriosis (DIE) are the three main clinical types. DIE is the most severe form and affects approximately 20% of all cases, defined as invading more than 5 mm in involved tissues (2).

Bowel is commonly involved in DIE, especially the rectum and the sigmoid colon, presenting as fibrotic nodules. Patients with bowel endometriosis can be asymptomatic or symptomatic. In addition to typically endometriosis-related symptoms, specific bowel symptoms can be observed, including constipation, diarrhea, tenesmus, etc. Medical intervention is not necessary for asymptomatic patients, but scientific management is required for symptomatic

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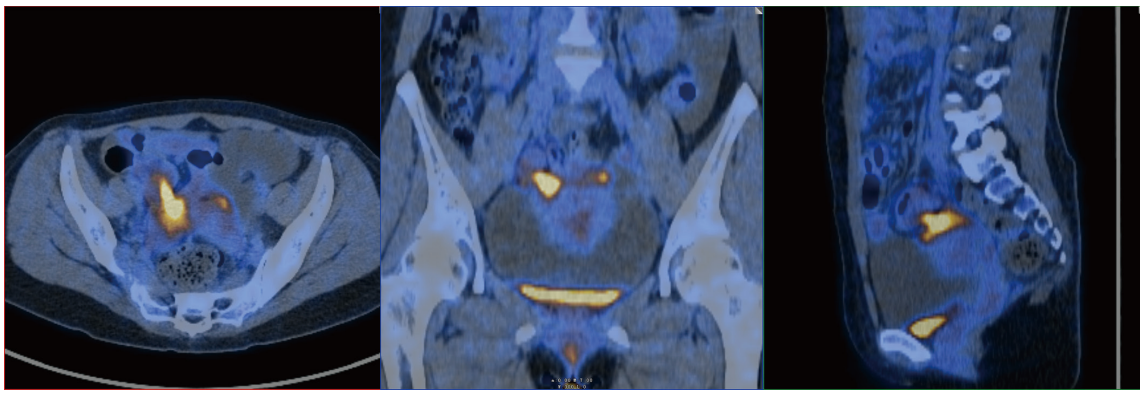

Figure 1 PET-CT revealed a right pelvic mass with an increased uptake of FDG. FDG, ^{18}F-Fluorodeoxyglucose; PET-CT, positron emission tomography-computed tomography.

population (3). Surgery is needed for patients with severe symptoms, failed hormone therapy or desired fertility. Surgical approaches of bowel endometriosis consist of shaving, disc excision and segmental resection. Each has its own indications, complications, and surgical outcomes (4). With individualized application, hormonal therapy after surgery may improve treatment outcomes (5).

Briefly, treatment of bowel endometriosis depends on many factors and individualized comprehensive management is still under exploration. Herein, an uncommon case of bowel endometriosis initially considered to be ovarian malignancy in the presence of pelvic mass and massive hemorrhagic ascites, treated by radical surgery and postoperative high-dose progestin without bowel recurrence, is reported to enrich clinical experience. We present the following article in accordance with the CARE reporting checklist (available at https://gs.amegroups.com/article/view/10.21037/gs-21-895/rc).

Case presentation

A 37-year-old, gravida 1 para 1 woman came to our hospital in June 2017 complaining of gradually worsen abdominal distention. She had a 10-year history of dysmenorrhea relieved with analgesics. Ultrasonography showed a right pelvic mass and massive ascites. A mildly elevation in CA-125 (126.9 U/mL, reference range <35.0 U/mL) was revealed in laboratory tests. Positron emission tomography-computed tomography (PET-CT) confirmed a soft tissue mass with an increased uptake of ^{18}F-Fluorodeoxyglucose (FDG) in the right pelvis (*Figure 1*). The patient was hospitalized for suspected ovarian malignancy.

A diagnostic abdominal paracentesis was performed, and 380 mL of hemorrhagic ascites was drained out. No malignant cells were found. Then she underwent diagnostic laparoscopy. A large amount of hemorrhagic ascites was identified upon entering the peritoneal cavity and 2 L was evacuated in total (*Figure 2A*). The pelvic anatomy was completely distorted by severe adhesions. The ileoceca was enlarged and rigid, forming a 5 cm × 5 cm mass (*Figure 2B*). Miliary nodules ranging from 0.5–1 cm in diameter scattered on the surface of the bowel (*Figure 2C*). Segmental bowel resection was performed to remove the ileocecal mass (*Figure 2D*). Other superficial or tiny lesions were removed by shaving and bipolar cautery. The pelvic anatomy was restored after complete adhesiolysis and there was no visible residual lesion. Both the intraoperative and postoperative pathology confirmed ileocecal endometriosis, and the resection margin was disease-free (*Figure 3*). After surgery, the patient immediately received 6-course gonadotropin-releasing hormone agonists (GnRH-a) therapy (leuprolide acetate 3.75 mg per course). To better prevent recurrence, two levonorgestrel intrauterine systems (each containing levonorgestrel 52 mg and releasing 20 μg/day) and two levonorgestrel subdermal implants (each containing levonorgestrel 75 mg and releasing 45 μg/day) were later inserted after multidisciplinary consultation and informed patient consent (6,7). After medication, she entered menopause without hot flashes, palpitation, or other discomforts.

The patient was asymptomatic without recurrence 25 months after surgery. In July 2019, she was readmitted for bilateral ovarian cysts found by pelvic magnetic resonance imaging (MRI). The second laparoscopy showed

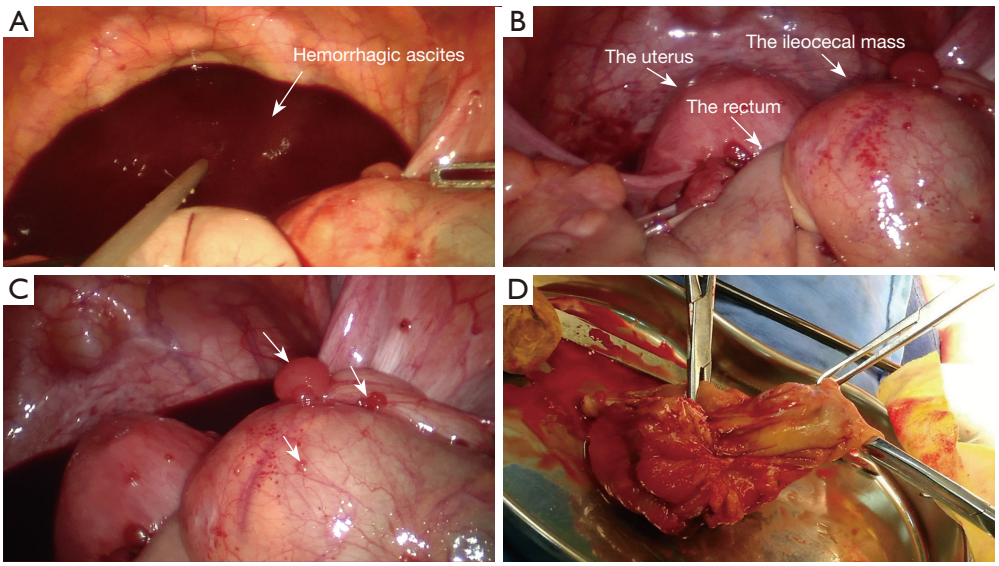

Figure 2 The first laparoscopy. (A) Massive hemorrhagic ascites; (B) severe pelvic adhesions; (C) miliary lesions on the bowel surface (arrows); (D) the ileocecal mass.

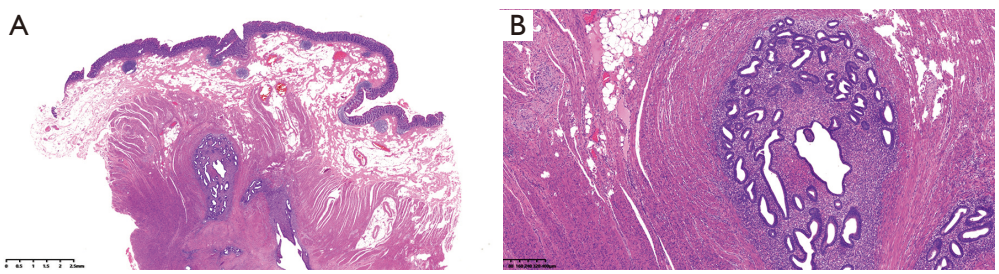

Figure 3 Hematoxylin and eosin staining of the ileocecal mass showed endometriotic glands infiltrated to muscularis (magnification ×1, magnification ×4).

the bilateral fallopian tubes were edematous, tortuous, and tightly adhered to the ovaries. The pouch of Douglas was completely enclosed by extensive adhesions (*Figure 4A*). However, the bowels were totally absent of recurrence (*Figure 4B*). Bilateral ovarian cystectomy and salpingectomy was performed (*Figure 4C*). Pathological examination demonstrated bilateral ovarian endometriomas. She completed 3-course GnRH-a therapy after surgery. On regular follow-up, no recurrence in bowels and no ascites were observed by November 2021.

All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

Discussion

Endometriosis presenting with ascites

Ascites related to endometriosis is uncommon. The first case was described by Brews in 1954, and to date less than 100 cases have been reported in publications (8).

When associated with ascites, endometriosis should be differentiated from ovarian malignancy. Cytological examination of endometriosis-related ascites shows hemosiderin-laden histiocytes instead of malignant cells (9). Core needle biopsy of thickened peritoneum has also been reported as a working method to confirm endometriosis (10).

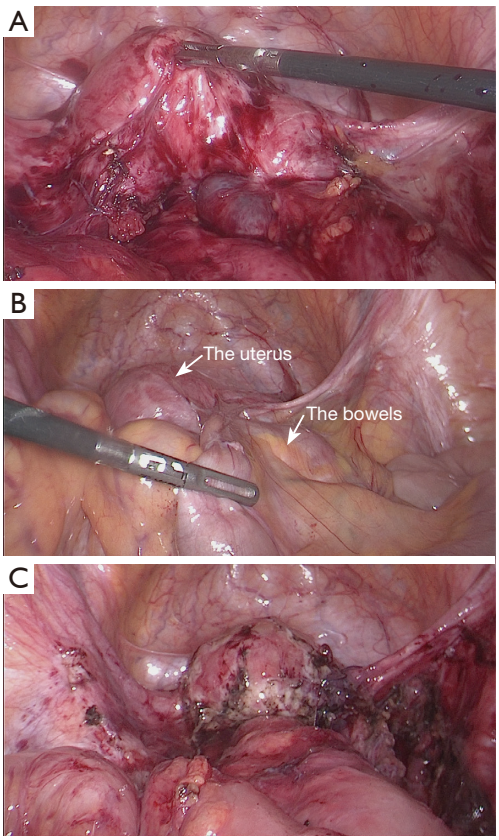

Figure 4 The second laparoscopy. (A) Pelvic adhesions; (B) no recurrence in bowels; (C) bilateral ovarian cystectomy and salpingectomy were performed.

However, in our case, despite negative ascites cytopathology, ovarian malignancy cannot be excluded without histological evidence, considering the elevated CA-125 and increased uptake of FDG.

Therefore, although these minimally invasive techniques are effective to clarify the source of ascites, diagnostic laparoscopy and histopathology are still the gold standards for endometriosis diagnosis and staging. Besides, gross ascites in the present case was reported to increase recurrence risk of endometriosis (8).

Surgical treatment for bowel endometriosis

Although hormonal therapy may improve symptoms, surgery is necessary for patients with severe symptoms, failed hormone therapy or desired fertility (3). There is an evident improvement of pain and other specific symptoms after surgery (4,11,12).

Surgical approaches are mainly divided into three categories: shaving, disc excision and segmental resection. Shaving means excising lesion without opening the intestinal lumen, usually indicated for rectal endometriosis. Disc excision is defined as a full-thickness resection of the bowel wall. Segmental resection means removal of a bowel segment with subsequent anastomosis, most frequently used for intestinal lesions (3).

An appropriate surgical technique depends on many factors, such as the size of lesions, depth of infiltration, extent of disease and distance from anal verge. In terms of size, only lesions smaller than 25–30 mm may be considerable for conservative surgery (shaving or disc excision) (13). The infiltrative depth is another consideration. Superficial lesions of serosa are suitable for shaving. When infiltration is deeper than muscular layer or shaving can cause significant bowel damage, disc excision is preferable. But at the same time, the lesion must be singular nodule, smaller than 3 cm, affecting less than 1/3 of the intestinal circumference (14). Segmental resection is required when multiple endometriotic nodules involve the same bowel segment, or bowel occlusion exceeds 50%, or nodule size is >3 cm (15). Compared with conservative surgery, a higher rate of severe complications was observed in segmental resection (3,16). Anastomotic leakage and rectovaginal fistula are the main complications (11). A temporary colostomy is advisable when lesions situate <6 cm from the anal verge (17).

In this case, different techniques were used for complete removal. The large ileocecal mass (5 cm × 5 cm) was removed by segmental resection and the superficial nodules were shaved. Other scattered tiny lesions in bowels and pelvis were electric burned, since they were too tiny to be shaved and extensive shaving may lead to bowel damage.

Adjuvant hormonal therapy

Endometriosis is of high recurrence rate, especially DIE (18,19). Immediately hormonal therapy after ovarian sparing surgery has been proposed for patients without fertility desire (5). A short-course therapy followed by long-period hormonal suppression is recommended because retrograde endometrium and ovulation in menstrual cycles may cause the formation of *de novo* lesions (20,21). Short-term GnRH-a has remarkable effects and the most used is six-month therapy (22). Progestins and combined oral contraceptives (COCs) are both effective and convenient for long-term administration. But COCs show more side effects and

contraindications than progestins. What's more, the estrogen contained in COCs may stimulate endometriosis (23,24).

The patient had multiple recurrence risk factors including bowel endometriosis, massive ascites, severe adhesions, and dysmenorrhea. She refused oral medication after surgery, but strongly desired for pain relief and recurrence prevention. Considering high-dose progestin contributes to recurrence prevention and dysmenorrhea relief (25,26), two intrauterine devices and subdermal implants were applied after multidisciplinary consultation and the total dose of levonorgestrel (130 μg/day) was reported to be safe (27).

Postoperative recurrence in ovaries

Although bowel recurrence was successfully prevented, it's worth discussing why new lesions appeared on ovaries. Continuous application of high-dose progestin leads to effective suppression of ovarian function indicated by anovulation and menopause. Under such conditions, there was little chance of forming *de novo* lesions. However, microscopic implants were reported to exist even when no visible residual lesion was observed (28). In this case, the regrowth of occult progestin resistant lesions after the first surgery may finally lead to the subsequent recurrence of ovaries.

Therefore, optimal surgical removal is of great significance and hormonal therapy may provide an additional component in the management of symptomatic bowel endometriosis.

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Footnote

Reporting Checklist: The authors have completed the CARE reporting checklist. Available at https://gs.amegroups.com/article/view/10.21037/gs-21-895/rc

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://gs.amegroups.com/article/view/10.21037/gs-21-895/coif). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

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