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Small Bowel Obstruction Caused by Ileal Endometriosis with Appendiceal and Lymph Node Involvement Treated with Single-Incision Laparoscopic Surgery: A Case Report and Review of the Literature

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

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Conflict of interest: None declared

Patient: Female, 40-year-old
Final Diagnosis: Intestinal endometriosis
Symptoms: Abdominal pain
Medication: —
Clinical Procedure: —
Specialty: Surgery

Objective: Challenging differential diagnosis

Background: Endometriosis is an ectopic proliferation of endometrial glands and interstitium outside the uterus. It usually affects the organs surrounding the uterus, and less often, involvement of extrapelvic organs, such as the intestines and urinary tract, is observed.

Case Report: A 40-year-old woman had been experiencing intermittent right lower abdominal pain for years, which worsened months earlier. The patient was admitted for the worst pain ever accompanying nausea and vomiting. Contrast-enhanced computed tomography revealed a heterogeneously enhanced lesion that measured approximately 50×25×35 mm, and a caliber change of the ileum at the same site with dilated small bowel proximal to the caliber change were observed. Colonoscopy revealed that the ileocecal valve and the lumen of the terminal ileum protruded inward, suggesting an extramural compression by the lesion. Since the patient showed no improvement following conservative therapy, bowel resection through a single-incision laparoscopic surgery was successfully performed. Histopathological exploration showed patchy infiltration of endometrium-like tissues in the muscularis propria and subserosa layers of the ileum and appendix. Moreover, nearby lymph nodes resected for their firmness showed similar findings.

Conclusions: We report a case of recurrent intestinal obstruction due to ileal and appendiceal endometriosis with lymph node involvement, which was successfully treated by single-incision laparoscopic surgery. Careful follow-up is important because the prognosis for the intestinal endometriosis with lymph node involvement is still unclear.

Keywords: Endometriosis • Intestinal Obstruction • Laparoscopy

Abbreviations: **CT** – computed tomography; **NPO** – nil per os; **SILS** – single-incision laparoscopic surgery

Full-text PDF: https://www.amjcaserep.com/abstract/index/idArt/930141

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Background

Endometriosis is a condition in which the endometrium, which originally constitutes the lining of the uterus, proliferates outside the uterus. It frequently occurs in the pelvis but affects the extrapelvic organs as well, including the intestines and urinary tracts [1]. Generally, endometriosis is treated with medication at first, and surgery is indicated when the initial conservative approach fails. Nevertheless, the intestinal endometriosis is mainly treated surgically due to the difficulty of preoperative diagnosis.

Here, we present a case of a patient who presented with recurrent episodes of small bowel obstruction, which had deteriorated over time. We treated the patient using a single-incision laparoscopic surgery (SILS) for its esthetic advantage and earlier recovery after surgery. Moreover, we describe this case with a literature review.

Case Report

A 40-year-old married woman, gravida 1 and para 1, had been experiencing intermittent right lower abdominal pain for years. During the 4 months before her visit to our hospital, the patient's pain gradually became incessant, and she visited a gynecologist at a nearby hospital, where she was diagnosed with right ovarian chocolate cyst as the origin of pain.

The patient finally visited our department for the worst pain ever accompanied with nausea and vomiting. On admission, the patient's height and weight were 170 cm and 53 kg, respectively. Her vital signs were as follows: blood pressure, 113/78 mmHg, heart rate 88 beats/min, and body temperature 36.7°C. The patient's abdomen was distended and soft with tenderness at the right lower quadrant without peritoneal signs. Laboratory results showed an elevated leukocyte count (13 200/μl) with a left shift. C-reactive protein was negative (0.02 mg/dl).

Abdominal contrast-enhanced computed tomography (CT) revealed a heterogeneously enhanced lesion, which measured approximately 50×25×35 mm (**Figure 1**). The caliber change of the ileum at the same site and dilated small bowel distal to the caliber change was observed. No dilated lymph nodes were found. A polycystic lesion was found in the right ovary, confirming the previously diagnosed right chocolate cyst. No other intrapelvic lesion or ascites was detected.

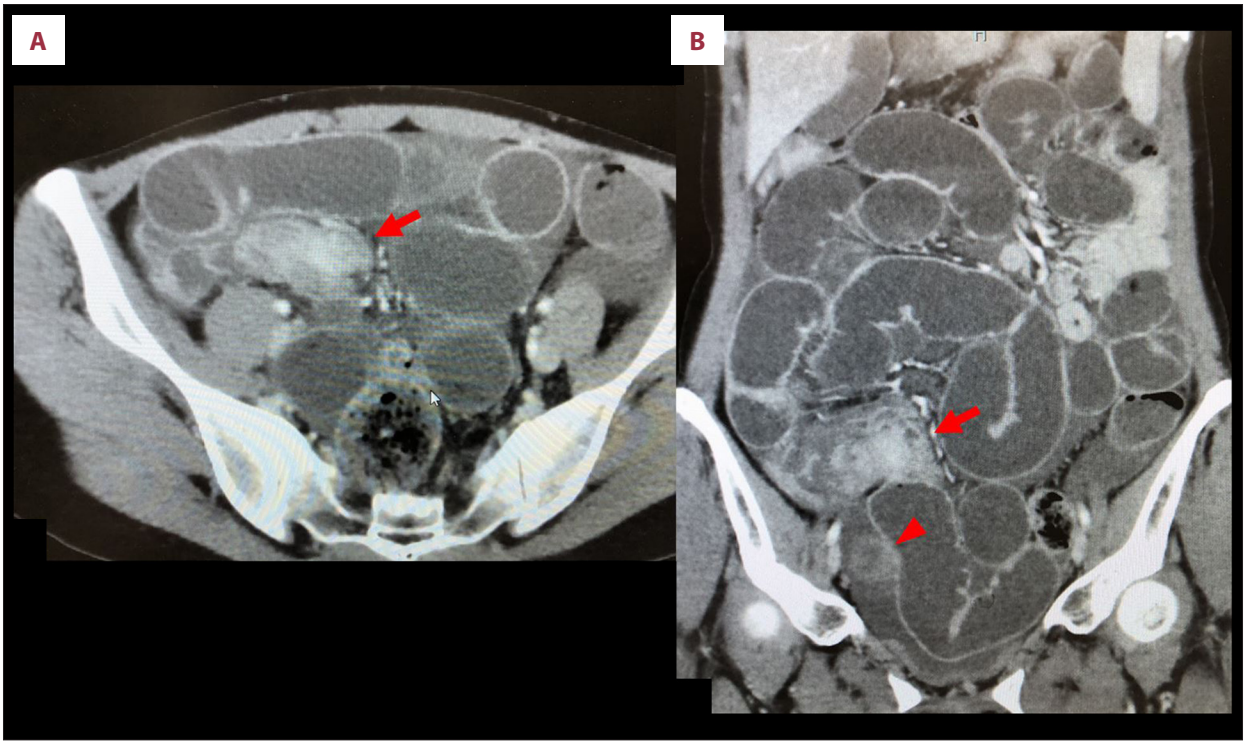

Figure 1. (**A, B**) Contrast-enhanced computed tomography (CT) findings. Abdominal contrast-enhanced CT revealed a heterogeneously enhanced lesion with cystic content, which measured approximately 50×25×35 mm (arrowhead). The caliber change of the ileum at the same site and dilated small bowel proximal to the caliber change were observed. No dilated lymph nodes were found. A polycystic lesion was found in the right ovary, confirming the previously diagnosed right chocolate cyst (arrow head). No other intrapelvic lesion or ascites was detected.

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Colonoscopy showed no epithelial lesion in the terminal ileum and the cecum, but the ileocecal valve and the lumen of the terminal ileum seemed to be rather protruded inward, suggesting compression by the extramural lesion (**Figure 2**). The appendiceal orifice could not be discerned due to the protruded ileocecal valve.

The patient was instructed to observe nil per os for the initial 3 days, but experienced no improvements; therefore, a nasointestinal tube was inserted on the fourth day. However, the patient's condition showed no improvement; therefore, surgical treatment was planned and performed on the seventh day.

Preoperatively, the differential diagnoses for this patient included Crohn's disease, intestinal endometriosis, diverticulitis with abscess formation, and intestinal tuberculosis. We presumed a malignant lesion was less likely considering the patient's background and clinical course.

We chose to perform a single-incision laparoscopic surgery (SILS) due to its advantages such as esthesis, less skin incision, and enhanced recovery after surgery.

The patient was placed in supine position and tilted to the left and head down to mobilize the small and large intestines to gain a good surgical field. An 8-cm incision at the umbilicus was made, and the EZ-access device with three 5-mm trocars attached was placed. Under CO_2 insufflation at a pressure of 8 mmHg, the laparoscopic intra-abdominal surveillance was performed. In the pelvis, serous ascites and the dilated right ovary were found (**Figure 3A**). No other lesion or adhesion was found in the pelvis. The lesion responsible for the bowel obstruction was easily found at the ileocecal region, forming a solid mass (**Figure 3B**). The mobilization of the ileocecal region using an inferior and lateral dissecting approach was performed using laparoscopic coagulating shears. After confirming that the ileocecal region could be pulled out from the incision line, the bowel resection and anastomosis were performed outside the abdomen. On observation, the small bowel obstruction was caused by a solid mass involving the appendix and terminal ileum. We scrutinized the entire length of the small intestine and found a small flat whitish lesion attached to the serosa, 6 cm proximal to the main lesion (**Figure 3C**). Thus, we decided to perform ileocecal resection including the whitish lesion (the length of the resected bowel was approximately 15 cm). The bowel was reconstructed by functional end-to-end anastomosis using linear staplers. Moreover, we resected lymph nodes along the ileocecal vessels, which were firmly palpable. We successfully completed the surgery with no postoperative complications, and the patient was discharged on the sixth postoperative day. The scar was minimal and esthetically satisfactory (**Figure 3D**).

Macroscopically, the resected specimen showed a solid mass that involved the terminal ileum and appendix (**Figure 4**). The lumen was clear, with no epithelial lesion. Histopathology showed patchy infiltration of glands forming similarly to tissues of endometrial glands and interstitium in the layers of the muscularis propria and subserosa of the appendix and terminal ileum (**Figure 5**). The white scar on the serosa of the ileum and the resected lymph nodes showed similar findings, indicating their involvement. No malignancy was observed.

Discussion

Endometriosis is a condition in which ectopic endometrium-like tissues proliferate in organs other than the uterus, forming solid adhesion to the invading tissue [2].

The etiology of endometriosis is still undetermined; however, 2 dominant theories exist regarding its origin. The most important hypothesis was proposed by Sampson in 1927, who observed menstrual bleeding flow back from the fallopian tube during operation, concluding that it causes implantation onto the nearby peritoneum [2]. The other is the coelomic metaplasia theory, explaining that the peritoneum directly causes metaplasia to become ectopic endometrial tissue on the grounds that endometriosis can occur even for patients who congenitally lack a uterus [3].

The occurrence rate of endometriosis in women of reproductive age is approximately 10% and still increasing [4]. The most affected site is the ovary, the pouch of Douglas, and the rectum, due its proximity to the fallopian tube, according to Sampson's theory. In contrast, endometriosis in the ileum and appendix is relatively rare [5].

Endometriosis affecting the appendix and/or ileum shows various symptoms, such as bowel obstruction, perforation, acute appendicitis, and intussusception [6-10].

However, the preoperative diagnosis is difficult since the symptoms, laboratory studies, and imaging studies are all nonspecific. Therefore, when women of reproductive age show chronic abdominal symptoms, endometriosis should be a differential diagnosis.

In this study, lymph node involvement was observed, suggesting its spread via the lymphatic system. Emerging evidence suggests that the lymphatic system is deeply involved in the pathogenesis and progression of endometriosis [11]. Previous studies revealed that gene expression levels of vascular endothelial growth factors (VEGFs) were altered in the uterine tissue of patients with endometriosis, indicating dysregulation of angiogenic and lymphangiogenic activity in the patients. Importantly, VEGF-C and VEGF-D, which are potent molecules for lymphangiogenesis, showed higher levels. Moreover, the

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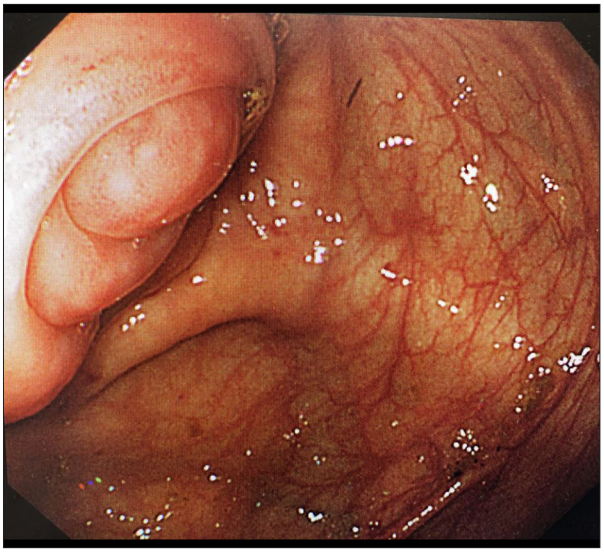

Figure 2. Colonoscopy findings. Colonoscopy showed no epithelial lesion in the terminal ileum and the cecum, but the ileocecal valve and the lumen of the terminal ileum seemed to be rather protruded inward, suggesting compression by the extramural lesion. The appendiceal orifice could not be discerned.

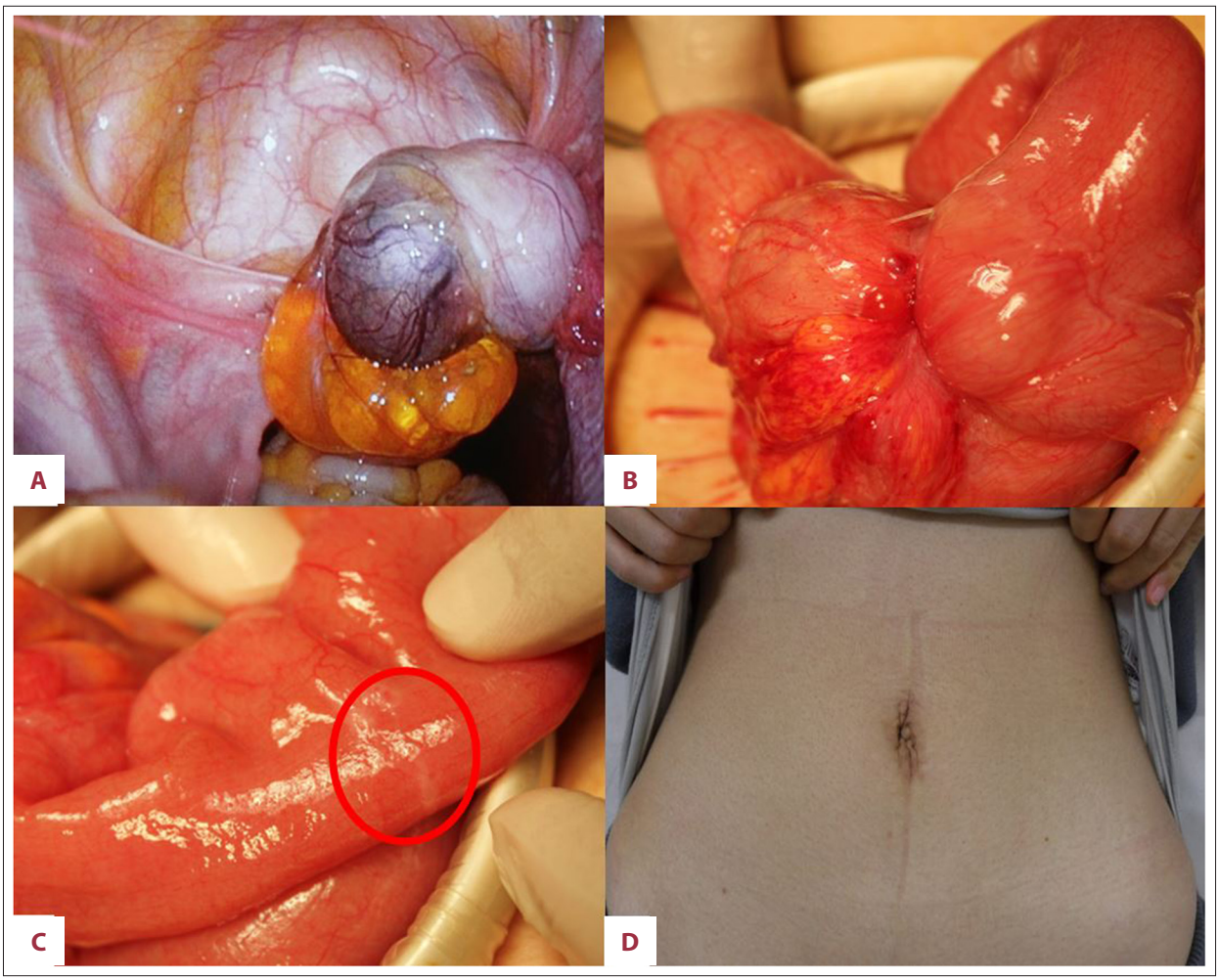

Figure 3. Intraoperative findings. The right ovary was enlarged. No other abnormalities were observed in the pelvis (**A**). The lesion responsible for the bowel obstruction was located at the terminal ileum, forming a solid mass (**B**). The small white lesion was attached to the serosa (**C**), which was resected together with the mass. The abdominal scar was minimal and aesthetically satisfactory (**D**).

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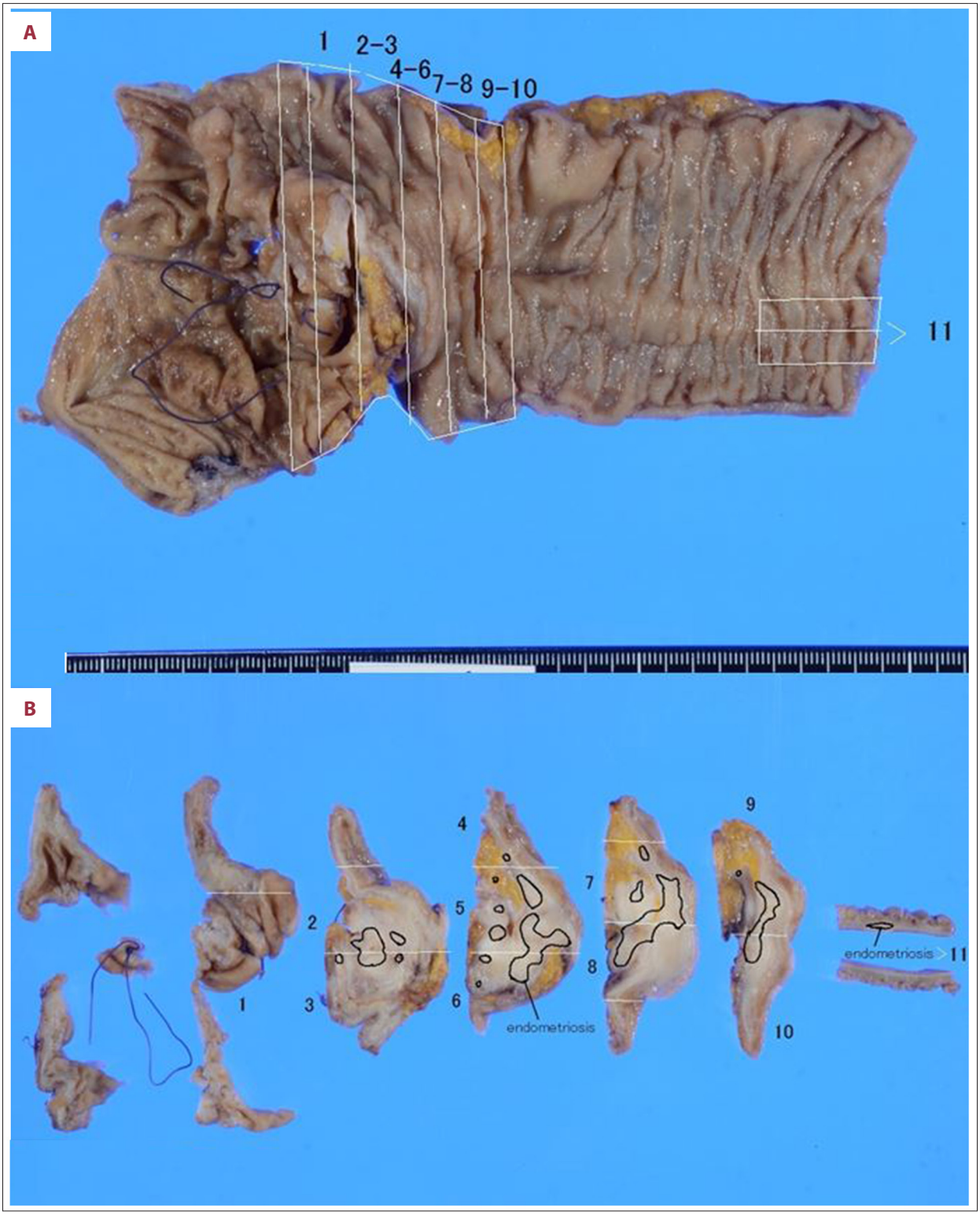

Figure 4. Macroscopic findings of the resected specimen. The resected specimen showed a solid mass that involved the terminal ileum and appendix. The lumen was clear, with no epithelial lesion (**A**). Histopathology revealed patchy distribution that showed infiltration of endometrial tissues (**B**).

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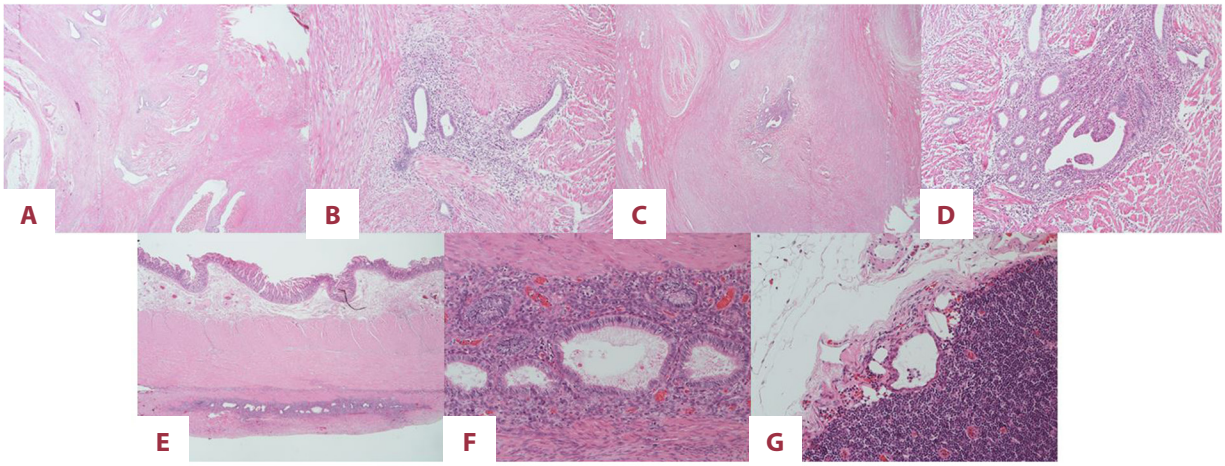

Figure 5. Histopathological findings. In the ileum (**A, B**) and appendix (**C, D**), gland formation having similarity with tissues of the endometrial glands and interstitium in the muscularis propria and subserosa was observed. The white scar on the serosa of the ileum (**E, F**) and the picked-up lymph nodes (**G**) showed similar findings (hematoxylin and eosin staining: **A, C, E:** ×20; **B, D, F, G:** ×100).

densities of lymphatic vessels are increased in ectopic endometriosis tissues, suggesting involvement of lymphatic spread in the pathogenesis of endometriosis [12-15].

The clinical impact of lymph node involvement of endometriosis remains unclear. In a study by Noël, 11 of 26 cases (42.3%) of rectosigmoid endometriosis showed lymph node involvement, which was associated with the size of the lesions and the presence of lymphatic and vascular involvements, indicating that it is probably more common than expected [16]. Rossini et al looked into the clinical and pathological correlation between the severity of endometriosis and lymph node involvement by studying 140 cases of colorectal endometriosis [17]. They found no correlations between the rate of intestinal stenosis, extent of infiltration, and the intestinal endometriosis recurrence rate. Intriguingly, considering that lymph node dissection is usually not performed, due to the benign nature of endometriosis, positive lymph nodes that were possibly left untreated have no clinical influence so far.

Generally, endometriosis is a progressive disease affecting women of reproductive age and causes chronic symptoms if untreated. In principle, ectopic endometrial tissues should be surgically removed to cure the disease [18]. Recently published guidelines developed by the Japan Society of Obstetrics and Gynecology and the Japan Society of Endometriosis indicate that surgical intervention can ease symptomatic intestinal endometriosis of the ileocecal region, small intestine, and appendix [19]. However, no reports have compared surgical treatment with nonsurgical treatments, suggesting that surgical treatment is recommended as an option when nonsurgical treatments fail.

Due to the difficulty in preoperative diagnosis, most cases of symptomatic gastrointestinal endometriosis are treated surgically. Therefore, as a more practical approach, accidentally found cases during surgery for other purposes may be preferably chosen to receive medical treatments, such as pain medication, nonsteroidal anti-inflammatory drugs (NSAIDs), and hormone therapy, including oral contraceptives and gonadotropin-releasing hormone analogs, which slow ectopic growth of endometrial tissues [20].

We chose to perform a single-incision laparoscopic surgery (SILS) to treat this case as the surgical approach. A recent meta-analysis has revealed that single-incision and conventional laparoscopic surgery had no difference in short-term clinical outcomes, such as mortality, morbidity, operative time, blood loss, conversion, perioperative complication, and hospital stay [21]. It is clear that SILS has an aesthetic advantage and causes less pain; therefore, it can be beneficial for patients with intestinal endometriosis who are relatively younger and do not need lymph node dissection.

Conclusions

We report the case of a recurrent intestinal obstruction due to appendiceal and ileal endometriosis with lymph node involvement, for which we performed a SILS. Careful follow-up is required because the prognosis for intestinal endometriosis with lymph node involvement remains undetermined. Regarding surgical procedure, SILS seems to be suitable, as it mostly affects women of reproductive age and it is a benign disorder.

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Conflict of Interests

None.

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