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A Report of Four Cases of Intestinal Endometriosis

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ABSTRACT

Four cases of intestinal endometriosis seen at our hospital are presented. The patients ranged in age from 35 to 43 years and developed abdominal pain, vomiting, and dyschezia due to stenotic lesions of the intestine. The sites of the lesions were the ileum in 2 cases, and the sigmoid colon and rectum in 1 case each. All cases had no history of bowel disease or laparotomy, and were not diagnosed preoperatively. These results suggested that evaluations of symptoms and clinical examinations are inadequate for an accurate diagnosis of intestinal endometriosis. The patients' postoperative courses were uneventful, and there have been no recurrences. In conclusion, intestinal endometriosis should be considered in women of childbearing age who present with bowel obstruction, especially in women without a history of laparotomy.

Key words: *Endometriosis, Ileus, Menstruating woman*

Intestinal endometriosis affects 12%-15% of menstruating women[3]. The range of symptoms of bowel endometriosis is wide, ranging from asymptomatic to a constellation of symptoms including painful bowel movements, cramps, constipation, diarrhea, vomiting, rectal pain, infertility, abdominal mass, increased urinary frequency, and cyclical hematochezia[10]. Classically, the symptoms become worse during menses, but this is not always the case. This myriad of symptoms makes the condition difficult to diagnose acutely and preoperatively. Thus, four rare cases of intestinal endometriosis that caused bowel stenosis are described. Three of the four cases reported required an exploratory laparotomy for diagnosis. This report serves as a reminder of this rare condition, as well as highlighting the diagnostic difficulties it can pose.

CASE REPORT

Case 1:

A 36-year-old woman with no significant past medical history was transferred as an emergency to our hospital because of abdominal pain and vomiting. The patient had no history of tuberculosis or other infectious diseases. There was also no history of radiation exposure, contact with poisonous chemicals, or genetic abnormalities. Her family had no history of bowel disease. On physical examination, there was rebound tenderness in the right lower quadrant of the abdomen. No abnormalities were found on gynecological examination. Laboratory tests showed remarkably elevated levels of C-reactive protein (CRP), with no other abnormalities. Computed tomography (CT) showed dilatation of the small intestines and bloody ascites in the pelvic space. These findings supported ileus, and the patient underwent emergency surgery, during which ileocecal bowel wall thickening and a luminal stricture in the distal ileum, resulting in proximal lumen expansion, were found. No other organs were involved. En bloc ileocecal resection was undertaken, and the histopathological examination confirmed ileal endometriosis. The patient was discharged on postoperative day (POD) 28. The patient received GnRHa hormonal therapy as an outpatient, and there has been no recurrence of the symptoms for 28 months after the surgery (Fig,1a-f).

Case 2:

A-43-year-old woman presented to our hospital with a 2-day history of worsening colicky central abdominal pain associated with distension and vomiting. She had no previous history of bowel disease. CT of the abdomen and pelvis was ordered to elucidate the cause. Her abdominal CT with

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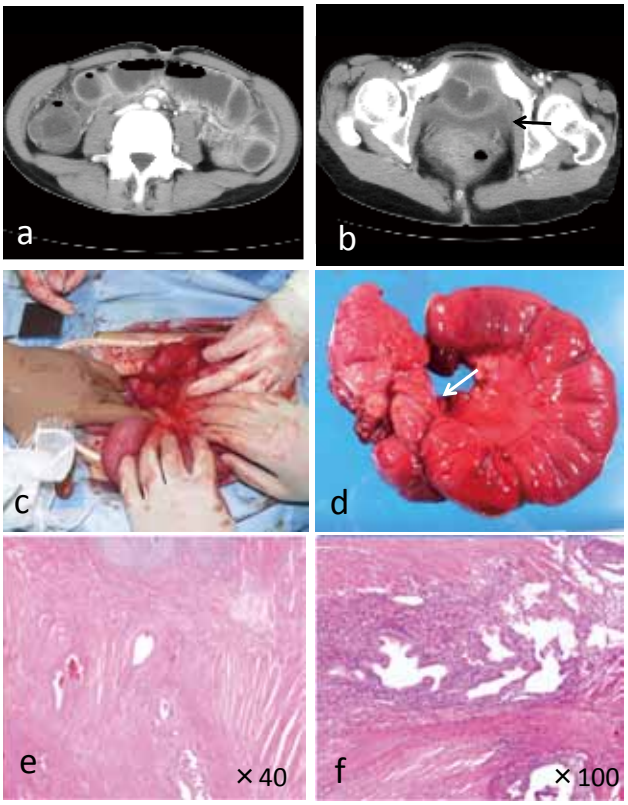

Fig.1: (a)(b) Abdominal CT shows dilatation of small intestine and ascites (black arrow) in the pelvic space. (c) A stenotic lesion detected in the terminal ileum at emergency surgery. (d) Macroscopic appearance shows stenotic lesion (white arrow) in the terminal ileum due to wall thickness. (e)(f) Microscopic examination confirms endometrial tissue in the serosal and muscular layers, with accompanying inflammatory cell infiltrations.

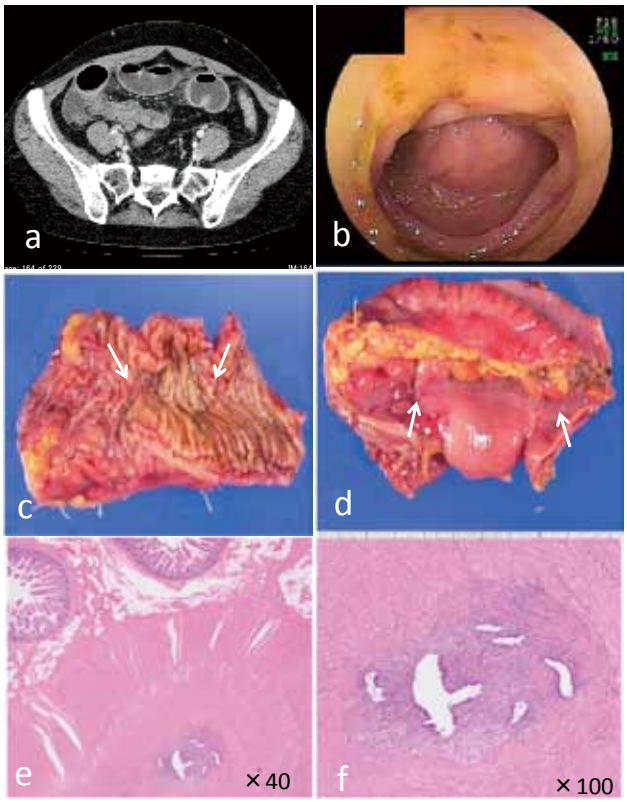

Fig.2: (a) CT scan shows dilatation and fluid retention in the distal ileum. (b) Endoscopy shows luminal stenosis of the ileum and no abnormal findings of the mucosal surface. (c)(d) Macroscopic appearance shows two stenotic lesions (white arrow) for several centimeters from Bauhin's valve. (e)(f) Microscopic examination confirms endometrial tissue in the muscular layer.

contrast performed on the admission day demonstrated small bowel dilatation with a transition point in the ileum, but no distinct mass lesion. The uterus and ovaries were unremarkable, and the adnexae were within normal limits. On endoscopy, luminal stenosis was seen at the ileum, 3 cm orally from Bauhin's valve, through which the endoscope could not pass, with no abnormal mucosa in the terminal ileum, indicating no evidence of pathological lymphadenopathy. These results showed frank small bowel obstruction with an area of focal nodular eccentric mural thickening. Laboratory tests showed markedly elevated CRP levels. No other abnormalities were found in tumor markers such as CEA or CA19-9. A partial resection of the small intestine was performed. Subsequent histopathology confirmed endometriosis of the distal terminal ileum causing two strictures. Postoperatively, the patient made an uneventful recovery and was discharged on POD 13. She was referred to a gynecologist for further management of her endometriosis. She received GnRHa hormonal therapy and was followed up at 8 months with no sign of recurrence (Fig.2a-f).

Case3:

A 35-year-old woman presented to our hospital with a history of lower abdominal pain and vomiting for the previous month that had been managed as gastroenteritis by her primary care physician. She was diagnosed as having endometriosis of the uterus and had been taking a low-dose contraceptive pill for 6 months. However, she interrupted the medication one month earlier. Laboratory data showed normal levels of CEA and CA19-9. A barium enema showed an elevated mass measuring approximately 3 cm in diameter in the sigmoid colon. Colonoscopy showed a submucosal tumor with a luminal stricture in the sigmoid colon and a longitudinal ulcer in the descending colon. Biopsy showed no evidence of malignancy or inflammatory bowel disease. She subsequently underwent laparoscopic sigmoidectomy, and histopathology confirmed intestinal endometriosis not involving the mucosa, with accompanying obstructive colitis. No subsequent medical treatment was given. The patient recovered well after surgery and was discharged on POD 22, and her quality of life has improved significantly. No recurrence of the symptoms occurred during 11 months of follow up. (Fig.3a-d).

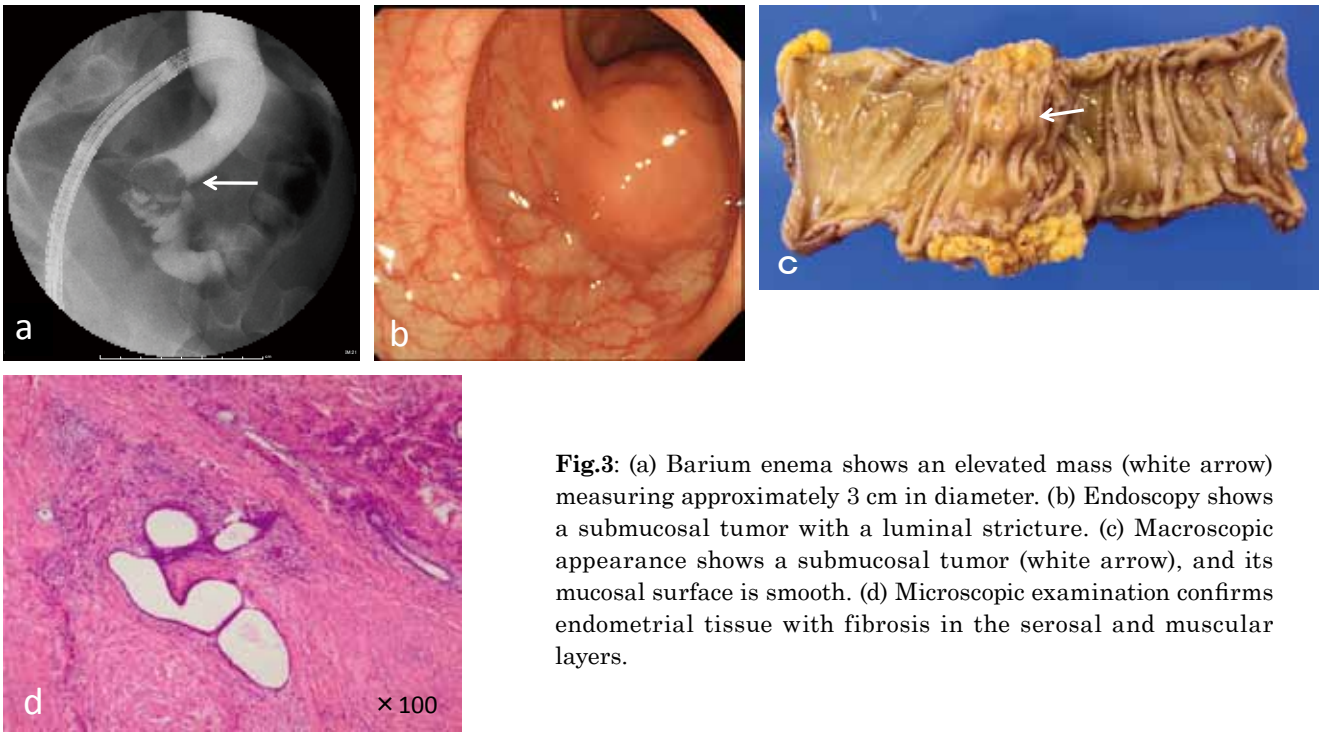

Fig.3: (a) Barium enema shows an elevated mass (white arrow) measuring approximately 3 cm in diameter. (b) Endoscopy shows a submucosal tumor with a luminal stricture. (c) Macroscopic appearance shows a submucosal tumor (white arrow), and its mucosal surface is smooth. (d) Microscopic examination confirms endometrial tissue with fibrosis in the serosal and muscular layers.

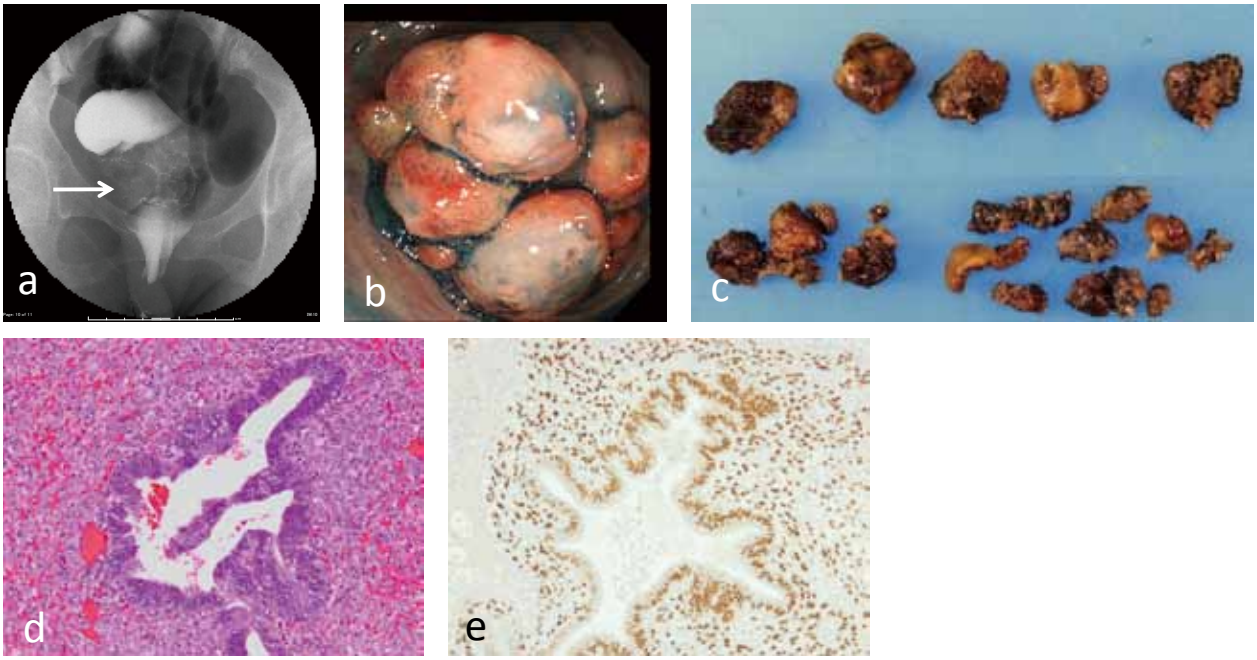

Fig.4: (a) Barium enema shows a semicircular rectal tumor (white arrow). (b) Colonoscopy identifies a lobulated nodular lesion with flare and erosion of the mucosal surface. (c) Macroscopic appearance of the excised rectal mass shows that it ranges in size from 0.5-3 cm. (d) Microscopic appearance confirms endometrial tissue in the submucosal layer. (e) Immunohistochemical analysis is positive for progesterone receptor.

Case 4:

A-35-year-old woman visited our hospital complaining of dyschezia and bloody stool for the previous year, and her endoscopic biopsy showed suspected mucosal prolapse syndrome (MPS). A barium enema showed a semicircular rectal tumor, and colonoscopy identified a lobulated nodular lesion with flare and erosion of the mucosal surface. Transanal excision was performed under a diagnosis of MPS using the transanal endoscopic microsurgery (TEM) method, and the diameter of the excised rectal mass ranged from 0.5-3 cm. Histological staining showed endometriosis in the submucosal layers, and immunohistological analysis showed positive staining for progesterone receptor in the nucleus. The patient received GnRHa hormonal therapy as an outpatient, and there was no recurrence of the symptoms in the 24 months after the surgery (Fig.4a-e).

DISCUSSION

Endometriosis is defined as the presence of ectopic endometrial tissue in extrauterine sites. It affects 10-15% of women of reproductive age and usually becomes apparent in the reproductive years when the lesions are stimulated by ovarian hormones[12]. Interestingly, although intestinal involvement is common in endometriosis, it rarely causes acute intestinal obstruction. The incidence of involvement of different intestinal sites varies greatly in the literature, with the rectosigmoid colon, small bowel, appendix and cecum affected in 50%-90%, 2%-16%, 3%-18% and 2%-5% of cases, respectively[16]. It was postulated by Lin et al, that this is due to the intestinal endometriosis being mainly an incidental finding[11].

The etiology of endometriosis remains unknown and controversial. There are many theories but currently the most widely accepted theory is that of retrograde menstruation causing the implantation and growth of endometriosis on the serosal surface of extra-uterine organs or occurring secondary to metaplasia in the pelvic peritoneum. The concept of retrograde menstruation is supported by the mainly pelvic distribution of endometriosis[6,13,14]. It is thought that the growth and invasion of endometriosis at ectopic sites are due to a process of neovascularization mediated by pro-angiogenic factors such as VEGF[15].

Small bowel endometriosis tends only to affect the bowel serosa. In rare circumstances, the disease can be more extensive; a histological review of 50 cases of intestinal endometriosis found that only 10% of intestinal cases had mucosal involvement[9]. Transmural disease damaging the mucosa can result in bleeding, the development of pseudo-tumors, or obstruction secondary to stenosis. In the present cases1,2, and 3, no mucosal change was detected, and histological staining showed endometriosis in the muscularis propria layers. On the other hand, in the present case 4, histological staining showed endometriosis in the submucosal layers, and colonoscopy identified the mucosal change. The strictures and masses arise from reactive smooth muscle hypertrophy secondary to disease present in the muscularis propria[9]. Rare cases of small and large bowel intussusception, bowel perforation, and malignant transformation have also been reported[1,8].

Acute bowel obstruction is a rare event occurring in less than 1% of cases of intestinal endometriosis and usually affects the rectosigmoid colon[5]. The two cases presented here are rarely seen, since small bowel obstruction accounts for only 0.7% of all surgical interventions for endometriosis[2]. As the present cases serve to highlight, in an acute presentation, the patient's history is unlikely to aid diagnosis, and, thus, it is unlikely for patients to be diagnosed preoperatively[7]. It is a challenging condition to diagnose because small bowel endometriosis can manifest with acute and chronic symptoms that can mimic many different pathologies, such as malignancy, inflammatory bowel disease, ischemic colitis, infectious disease, and irritable bowel syndrome (IBS)[7,9]. Hematochezia is also an uncommon symptom due to the low incidence of mucosal involvement[7]. The symptoms of intestinal endometriosis can be associated with the patient's menstrual cycle in 18-40% of cases[7]. However, without a high index of suspicion these symptoms may not be elucidated or considered important, particularly in an acute setting. This was clearly seen in the present cases 1,2, and 4, where the patients had no history of endometriosis and could not have been diagnosed preoperatively.

Contrast studies such as a barium enema may be helpful, although they are falling out of favor and may not be specific. Endoscopy may provide no valuable results because of the intact mucosa, but it is still recommended in all patients with suspected endometriosis to rule out mucosal involvement and malignant lesions with the help of biopsies, if needed. As was evident in the present cases 1, 2, and 3, multislice CT may also not be helpful, since it can be difficult to distinguish between ileal endometriosis and Crohn's disease, although it can be useful in diagnosis because it may demonstrate focal or constricting bowel disease. These findings, therefore, suggested that it is rare to be able to rely solely on imaging for the diagnosis of intestinal endometriosis.

Medical treatment with hormonal therapy such as the oral contraceptive pill (OCP), danazol, or gonadotrophin antagonists can be attempted for intestinal disease when there is no obstruction. This remains controversial because there have been few reported cases of medical therapy being successful[3]. Indeed, in Case 3 the patient's use of the OCP seemed to have some bearing on the progression of the disease, because the cessation of the OCP induced abdominal pain and vomiting. In addition, it is argued by some physicians that the rare but potential risk of malignant transformation makes surgical resection mandatory[3]. Surgery is only indicated in acute or subacute bowel obstruction that fails to resolve, as well as in endometriotic tumors or when it is impossible to exclude malignancy[7,8]. In an emergency setting, the main aim of surgery should be to relieve the obstruction[3]. Postoperative hormonal therapy is advocated by some physicians, but meta-analysis has failed to demonstrate any benefits[3,4]. In the present cases three of the four patients had cooperated with the gynecologist and received postoperative hormonal therapy for disease control and prevention.

In conclusion, acute bowel obstruction secondary to intestinal endometriosis remains a difficult condition to diagnose without an elevated index of suspicion. Therefore, intestinal endometriosis should be borne in mind when assessing women of repro-

ductive age who present with a small or large bowel obstruction, especially without a history of laparotomy. A careful history may elicit symptoms related to the patient's menses. CT and endoscopy are still recommended in all patients to rule out mucosal involvement and malignant lesions. In all of the present cases, the final diagnosis was made by the pathologist's postoperative report. Multidisciplinary care should be encouraged to ensure correct evaluation and improve the management of these patients.

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