# Distal Ileal Endometriosis as a Cause of Ileus: A Case Report

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**SUMMARY**

**Introduction** Endometriosis is a benign condition affecting females of reproductive age. Although intestinal endometriosis is common, it is rarely manifested as an acute bowel obstruction secondary to ileal endometriosis. Enteric endometriosis should be considered as a differential diagnosis when assessing females of reproductive age with acute small bowel obstruction.

**Case Outline** A 41-year-old woman presented with symptoms and signs of an acute small bowel obstruction requiring emergency surgery. A small bowel resection was performed with end-to-end anastomosis. Histological examination demonstrated endometriosis with fibrosis and stricture of the ileal segment. This case is important to report as it highlights the diagnostic difficulty this particular condition presents to an emergency surgeon.

**Conclusion** In the differential diagnosis, endometriosis should be taken into consideration when assessing females of reproductive age who present with abdominal pain and small bowel obstruction.

**Keywords:** intestinal endometriosis; acute intestinal obstruction; emergency surgery

## INTRODUCTION

Endometriosis is a chronic, benign, estrogen-dependent condition characterized by the presence of endometrial tissue outside the uterine cavity. This ectopic endometrium has the same histological characteristics like the normal one. Under the influence of estrogen endometriosis implants may proliferate, forming endometriosis nodules or cysts (endometriomas) and rarely infiltrate other organs [1, 2].

Endometriosis is a common finding in women aged between 30-40 years, and the lesions are stimulated by ovarian hormones [3, 4]. Endometriosis usually affects the genital tract; on the second place is the gastrointestinal (GI) tract with 3-37%, followed by the urinary tract with 10% of all women with this condition [5, 6]. Endometriosis rarely affects extra abdominal organs, such as lungs, skin and the central nervous system [7]. Extra-pelvic endometriosis affects the GI tract; the incidence of the involvement of different intestinal sites varies greatly in the literature with the rectosigmoid affected in 50-90%, small bowel 2-16%, appendix 3-18%, and caecum in 2-5% of cases [8]. Intestinal involvement in endometriosis is common but it rarely causes acute intestinal obstruction [8]. Although Sampson [9] first described the aetiology of endometriosis 90 years ago, it still remains a very complex clinical entity with non-specific symptoms which make it difficult to establish the diagnosis.

We report a case of acute small bowel obstruction secondary to distal ileum endometriosis requiring emergency surgery. The diagnosis of ileal endometriosis was made by histological examination of the resected ileum.

## CASE REPORT

A 41-year-old woman was admitted at our Clinic for Emergency Surgery with acute abdominal pain, nausea and vomiting. The patient was complaining of a three-day history of abdominal pain. On the day of admission the pain was localized in the epigastrium and was afterwards spread through the whole abdomen. The patient's past medical history included one uncomplicated Caesarean section, laparascopic removal of a left ovarian cyst 6 years ago when the endometriosis was histopatologically diagnosed. Two years after the surgery an evaluation by her gynaecologist diagnosed a right ovarian cyst that withdrew by hormonal therapy. She was taking antihypertensive therapy at the time of admission. Last period was two years ago. There was no family history of hereditary diseases. She was a non-smoker and did not drink alcohol. On examination the patient was comfortable at rest, haemodynamically stable, body temperature was 37.5°C.

Physical examination showed a distended abdomen with diffuse tenderness, most notably to the right of the umbilicus and right hypogas-

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trium. No pathological abdominal masses were found. Auscultation detected weakening peristaltic rushes. She had no adnexal tenderness. Rectal examination was regular.

Laboratory tests showed leucocytosis ($12.7\times10^9$). Other laboratory tests were normal. Ultrasound imaging showed free liquid in the Douglas space. Abdominal radiograph showed dilated loops of the small bowel, without pneumoperitoneum and colon distension. The patient was treated conservatively with intravenous rehydration therapy and nasogastric suction. A few hours after admission at our Clinic the patient redeveloped signs and symptoms of obstruction with diffuse abdominal pain and vomiting. The abdomen was distended without peristaltic rushes. Because of rapid deterioration the patient's condition with clinical and radiographic signs of obstruction surgery was immediately performed. After adequate perioperative preparation we made explorative laparatomy and found 2 litres of transparent ascites with diffuse dilation of the small bowel. Twenty centimeters proximally from the Bauchini valve we saw a fibrose stricture which occluded the bowel lumen. Proximally from the occlusion the bowel loops were oedematose and suculent. We evacuated ascites and sent it for citological examination, which was negative. Ileum resection was performed 5cm in length with a termino-termino EE anastomosis. Diagnosis was made by histological examination. Histology of the resected specimen showed endometriosis involving the distal ileum (Figures 1 and 2).

The patient had uneventful post-operative recovery and left the hospital 6 days later.

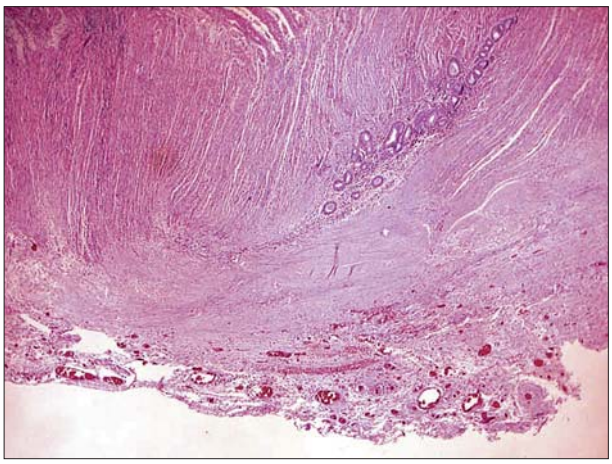

**Figure 1.** The sample of ileal wall which showed fibrostenotic area on gross examination, histologically revealed irregular tubulo-glandular arborization with endometrial epithelial lining and variability cellular and partly hyalinized stroma

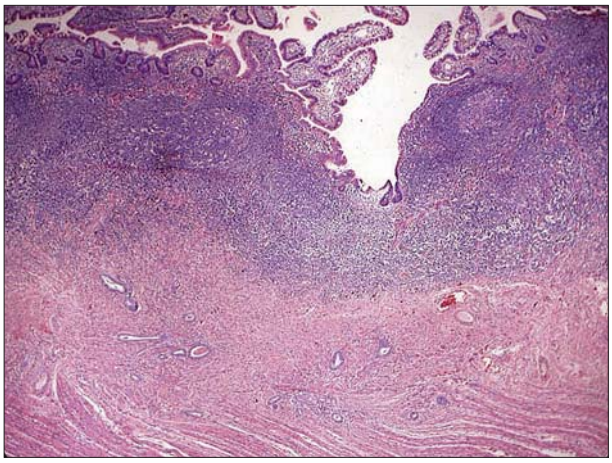

**Figure 2.** Endometriotic tissue infiltrates ileal submucosal layer beneath the hyperplastic mucosal lymphoid tissue

## DISCUSSION

There are more than 10 million women with endometriosis, 30% of amenorrhoeic women have endometriosis and the incidence of endometriosis is 30-50% in infertility couples [10, 11]. The reported incidence of the involvement of different intestinal sites varies greatly in the literature; this is due to intestinal endometriosis being mainly an incidental finding [8]. In a retrospective study of 7,000 patients with endometriosis the incidence of caecal and appendix involvement was 4% and 3% [12]. A similar result was shown in a study by Chapron et al. [13] assessing the anatomical distribution of endometriosis; appendix and ileocaecal involvement was found in 6.4% and 4.1% of intestinal cases, respectively.

The aetiology of endometriosis remains controversial. Many theories have been proposed to explain this condition, such as transformation of pluripotential peritoneal mesothelium, neurological hypothesis and migration of cells through the lymphatic system or via haematogenous spread [14, 15]. Immunological, genetic factors and unknown environmental factors could be involved in the pathogenesis of this disease [16]. It is thought that the growth and invasion of endometrial tissue at ectopic sites is due to a process of neovascularisation mediated by proangiogenic factors such as VEGF [17]. The most widely accepted theory is the Sampson's retrograde menstruation theory; during menstruation the endometrial tissue refluxes through the fallopian tubes, implanting and growing on the serosal surface of abdominal and pelvic organs [9]. The theory is supported by the mainly pelvic distribution of endometriosis [13].

Small bowel endometriosis tends to affect only the bowel serosa and deposits tend not to be larger than 2 cm in size; it is characterized by a patchy involvement of the bowel and macroscopically is glistening grey in appearance [8, 18, 19]. Histological examination of small bowel endometriosis shows a gradually spreading lesion from serosa to muscularis propria [14]. On the other hand, a rare but potential risk of malignant transformation makes surgical resection mandatory [20]. Serosa is rarely affected, because of its weak innervation [21]. Lymph node involvement of endometriosis can be affected as a consequence of lymphatic endometrium dissemination from the intestinal wall [22]. Although bowel endometriosis is generally asymptomatic, infiltration of muscularis propria can lead to local inflammation resulting in fibrosis and the formation of adhesions [5, 18]. Acute bowel obstruction is a rare event occurring in less than 1% of intestinal endometriosis and usually affects the rectosigmoid colon [23]. Small bowel obstruction has been found in only 0.7% of all sur-

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gical interventions for endometriosis [23]. Distal ileum obstruction occurs in 7-23% of all cases with intestinal involvement [23]. Here we present a rare case of small bowel obstruction requiring emergency surgery. Histology of the resected specimen showed bowel fibrosis caused by endometriosis. Rare cases of small and large bowel intussusception, bowel perforation and malignant transformation through adenocarcinoma have also been reported [24, 25, 26]. Malignancy has been reported in less than 1% of patients and 79% of these cases occur in the ovary, while only 1/3 of all cases is extragonadal [20, 27].

The diagnosis of small bowel endometriosis may be difficult because of different acute and chronic nonspecific symptoms. Clinical signs of endometriosis can mimic different pathological conditions, such as inflammatory bowel diseases, Crohn's disease, acute appendicitis, diverticulosis, infectious diseases, ischemic enteritis, and tumours [8, 24, 28, 29]. As we can see, the patient was admitted at our hospital with nonspecific abdominal symptoms which later redeveloped signs and symptoms of obstruction.

The most common symptom of enteric endometriosis is a colicky abdominal pain, which is a non-specific symptom in cases of bowel obstruction [3, 8]. Other common symptoms, such as nausea, emesis, pyrexia, constipation, weight loss and anorexia in isolation have a low diagnostic validity [8]. Haematochesia is an uncommon symptom due to the low incidence of mucosal involvement [3, 24]. Rarely, intestinal endometriosis may occur with perforation [25, 30].

There are many techniques in diagnosing intestinal endometriosis, such as double contrast barium enema, transvaginal ultrasonography, rectal endoscopic ultrasonography, magnetic resonance imaging and multislice computed tomography (MSCT) enteroclysis [28, 31]. In the case of endometriosis abdominal ultrasound and abdominal radiograph offers insufficient and often non-specific information, but bowel obstruction can be found. MRI is currently the best imaging modality for enteric endometriosis with sensitivity of between 77-93% [18, 28]. In our case the patient had increased inflammatory markers with non-specific ultrasound imaging and abdominal radiograph showed dilated loops of the small bowel. Because of rapid deterioration of the patient's condition an emergency laparatomy was immediately carried out and other imaging tests (MSCT, MRI) could not be achieved prior to surgery.

In the case of elective surgical treatment a laparoscopic approach is the gold standard [18, 19]. Surgery is indicated in acute or subacute bowel obstruction, endometriotic tumours, or when it is impossible to exclude a malignancy in cases of progressive pain and bleeding. In an emergency setting the main aim of surgery is to relieve the obstruction [1, 10, 24].

It is important to know that endometriosis has some unique biological characteristic; it is a chronic recurrent disease because of microscopic implants which are active without surgery [10]. If endometriosis is suspected intra-operatively, then as many ectopic deposits as possible should be excised [18]. In the case of the presence of bowel lesion a resection margin greater than 2 cm should be attempted [24]. If it is difficult to exclude a malignancy intra-operatively, it is appropriate to carry out oncological resection [8].

The case of small bowel obstruction presented here shows that endometriosis remains a challenging condition for clinicians. In the differential diagnosis, endometriosis should be taken into consideration when assessing females of reproductive age who present with abdominal pain and small bowel obstruction. Surgical treatment is necessary with the main aim to excise the point of obstruction and all deposits. Exclusion of bowel malignancy is essential and, if in doubt, oncological resection should be performed.

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# Ендометриоза дисталног илеума као узрок опструкције црева – приказ болесника

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**КРАТАК САДРЖАЈ**

**Увод** Ендометриоза је хронично бенигно обољење које се јавља код жена у репродуктивном периоду. Иако је она релативно честа дијагноза, ретки су случајеви акутне опструкције танког црева изазваних ендометриозом. У тим ситуацијама увек треба размишљати о ендометриози као диференцијалној дијагнози.

**Приказ болесника** Код четрдесетједногодишње жене јавили су се симптоми опструкције танког црева, те је она хитно оперисана. Ресекција танког црева је урађена по типу термино-терминалне анастомозе. Хистопатолошки преглед је указао на ендометриозу са фиброзом и сужењем дела илеума. Ендометриоза танког црева није тако честа дијагноза. Приказани случај указује на бројне дилеме с којима се хирург сусреће током дијагностичког процеса.

**Закључак** Треба размишљати о ендометриози као диференцијалној дијагнози код жена у репродуктивном периоду с клиничком сликом акутног бола у абдомену и опстукције танког црева.

**Кључне речи:** цревна ендометриоза; акутна опструкција црева; хитна лапаротомија

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