

## Case Report

## Puerperal ileal perforation secondary to endometriosis: Case report and literature review



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## ABSTRACT

**Objective:** Bowel endometriosis is an uncommon disease that can cause serious complications and may require immediate medical attention. We wish to remind about bowel perforation caused by endometriosis, its diagnostic difficulty, and the need or urgent management in late pregnancy and puerperium. **Case Report:** We present a 38-year-old woman, which presented with bowel perforation requiring urgent surgery. A pathological exam disclosed deep ileal infiltrative endometriosis.

**Conclusion:** Even though bowel endometriosis is a rare complication, it should be considered in the differential diagnosis of severe abdominal pain in late pregnancy or puerperium. A multidisciplinary management of these patients is needed.

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## Introduction

Endometriosis is the presence of endometrial glands or stroma outside the uterine cavity [1]. It mostly affects women of child-bearing age [2].

Symptoms usually appear during menses, due to their hormone-dependent nature. Dysmenorrhea, chronic pelvic pain, infertility, dyspareunia, and urinary and bowel disturbances may be present.

The prevalence of intestinal endometriosis ranges between 5.3% and 12%. The rectum and sigmoid colon are most commonly involved, while the ileum is rarely involved (4.1%) [3]. The average age at diagnosis is 34–40 years [4].

Differential diagnosis includes irritable bowel syndrome, infectious diseases, ischemic enteritis, Crohn's disease, and neoplasm [3].

Since there are no pathognomonic signs and symptoms, the preoperative diagnosis of bowel endometriosis is difficult [4].

We present a case of deep terminal-ileum endometriosis, which presented as perforation and peritonitis in early puerperium.

## Case Report

The patient was 38 years old. Her brother had died at the age of 54 because of colon cancer, and her sister suffered premenopausal breast cancer. The patient did not have remarkable antecedents, and denied genital endometriosis symptoms. A prior delivery was uneventful.

At 21 + 1 gestational week (November 2012), she was referred because of severe anemia (hemoglobin 3.9 g/dL). The rest of the laboratory tests and the regular pregnancy checks had been normal. The patient described epigastralgia and melena. She denied ingestion of nonsteroidal anti-inflammatory drugs.

Tests for hepatitis A, B, and C viruses, and human immunodeficiency virus; gastric lavage; and a colonoscopy were unremarkable. There was fecal occult blood (1723 ng/mL; normal value < 74 ng/dL). An upper gastrointestinal endoscopy was performed, and the biopsy of the second portion of the duodenum disclosed slight villous atrophy without lymphocyte infiltration.

The first diagnosis was celiac disease, and the patient clinically improved. Normal vaginal delivery took place in March 2013.

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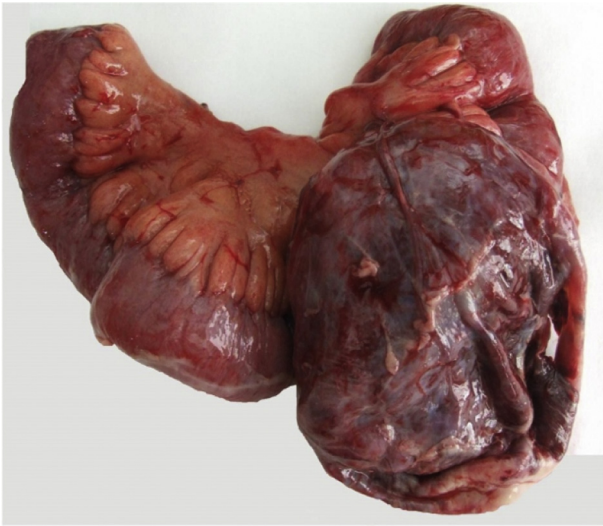


Figure 1. Surgical specimen.

The terminal ileum (30 cm), omentum, and peritoneal implants were resected (Figure 1), and peritoneal fluid samples were taken for cytological study and culture.

The pathology disclosed a deep nonpolypoidal infiltrative endometriosis of the small bowel (Figure 2), with vascular and lymphatic invasion, and subcapsular lymph-node involvement (Figure 3). The immunohistochemical study was positive for estrogen and progesterone receptors, CD10, and vimentin, whereas it was negative for CKAE1-AE, inhibin, C-KIT, CD34, desmin, caldesmon, and specific muscular actin (HHF35).

The initial postoperative course was uneventful until the 6<sup>th</sup> day, when rectal bleeding appeared and the hemoglobin dropped to 8.3 g/dL. The arteriography did not reveal active bleeding or abnormalities amenable for embolization therapy. Gastroscopy and colonoscopy were normal. Dynamic gammagraphy with Tc 99-marked erythrocytes showed proximal jejunal bleeding.

The patient was scheduled for quarterly gonadotropin-releasing hormone (GnRH) agonists. After the second dose of GnRH, she was started on oral contraceptives, remaining asymptomatic.

## Discussion

Endometriosis is a disease that affects between 10% and 15% of women at reproductive age [3]. Although it is considered a benign disorder, it can sometimes have an aggressive behavior, involving visceral lymphatic vessels and lymph nodes, and causing other serious complications that may require immediate treatment.

Intestinal endometriosis is the most common extrapelvic location; nevertheless, its prevalence is unknown. Most studies describe prevalence between 5.3% and 12%, but rates as high as 37% have been reported [2,5].

Intestinal endometriosis commonly affects the serosal and muscular layers of the bowel [2], while transmural involvement into the mucosa is rare. *Deep bowel endometriosis* is defined as a solid mass situated deeper than 5 mm under the peritoneum [6]. Mucosal involvement is the most severe form of bowel endometriosis. The most frequent locations are the rectum and sigmoid colon, whereas involvement of the small bowel (2–16%), appendix (3–18%), cecum (2–5%), and ileum (4.1%) is exceptional.

At postpartum, an endoscopic capsule study was performed. Only extrinsic compressions were described, with normal mucosa from the jejunum to the ileum, without bleeding.

Five months after delivery, during breastfeeding (August 2013), the patient arrived to the emergency room complaining of severe abdominal pain, with signs of peritonitis. The uterus and ovaries appeared normal with the transvaginal ultrasound. A cystic-appearing lesion was found in the right lower quadrant compartmental, with dense content. Computed tomography revealed a predominantly cystic mass located in the pelvis, suggesting an aneural origin. Multiple implants in mesenteric fat, ascites, as well as an enlarged left iliac lymph node suggested an ovarian cancer with peritoneal carcinomatosis.

An urgent laparoscopic surgery was performed. Peritonitis, a 5-cm solid tumor located in the terminal ileum and an adjacent cyst 21 cm in diameter, as well as parietal peritoneal and omental implants were found. The internal genitals were normal.

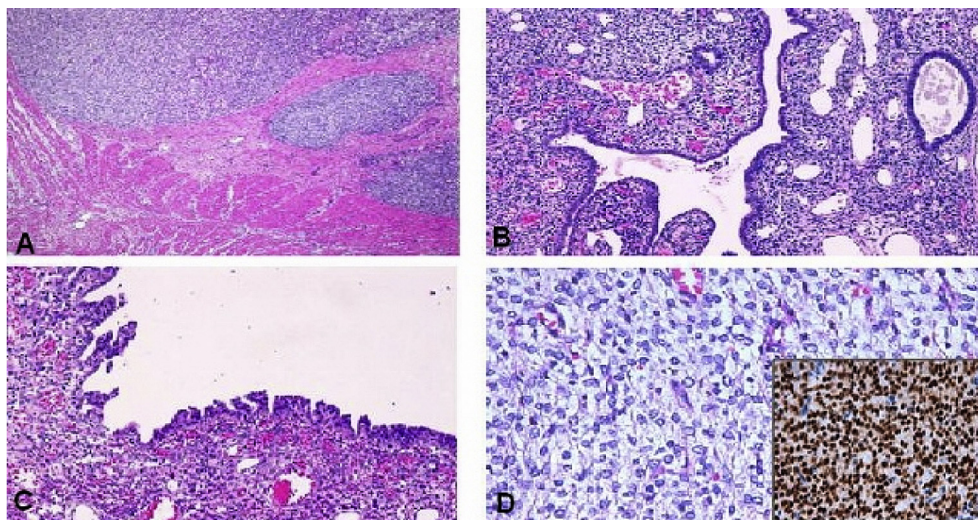


Figure 2. (A) The lesion infiltrated the muscular layer of the small bowel and showed a lobular arrangement. (B) It is consisted of dilated endometrial glands and stroma. (C) The glands were lined by a single layer of cuboidal epithelial cells with eosinophilic cytoplasm. (D) Stromal component consisted of small bland ovoid-shaped cells in an abundant myxoid matrix. There was no nuclear pleomorphism, and isolated mitotic figures were identified. Immunohistochemical staining shows diffuse and strong nuclear immunoreactivity of estrogen and progesterone receptors.

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