

## Case Report

# Intestinal Obstruction Secondary to Cecal Endometriosis

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## INTRODUCTION

Endometriosis refers to the presence of endometrial glands and stromal tissue domiciled primarily outside of the uterine corpus.<sup>[1,2]</sup> It can be broadly divided, based on the site of the lesion, into pelvic and extrapelvic endometriosis.<sup>[3]</sup> The former refers to endometriosis of the pelvic peritoneum, ovaries, and fallopian tubes,<sup>[4]</sup> while the latter refers to endometrial glands and stromal implants found in other areas of the body, of which the gastrointestinal tract is the most common site.<sup>[3]</sup>

Endometriosis is diagnosed in 1% of all patients undergoing major gynecological surgeries.<sup>[5]</sup> The reported incidence of endometriosis in asymptomatic women, infertile patients, and women with chronic pain has been reported as 1%–20%, 10%–25%, and 60%–70%, respectively.<sup>[6]</sup> Of all women of childbearing age with endometriosis, bowel involvement has been reported in 3%–37%.<sup>[5-7]</sup> Of these, the rectosigmoid, the small

## ABSTRACT

Endometriosis of the gastrointestinal tract is rare and occurs in <1% of all patients undergoing major gynecological surgeries. Bowel involvement has been reported in 3%–37% of all women of childbearing age with endometriosis. Total obstruction of the gastrointestinal tract occurs in <1% of cases of endometriosis with bowel obstruction. This case report is that of a 42-year-old female who presented with a 6-month history of change in bowel habits in favor of increasing constipation. This was associated with cyclical lower abdominal pains, abdominal swelling, and weight loss. Examination revealed hyperactive bowel sounds with scant fecal matter on rectal examination. An impression of intestinal obstruction was made and she had an emergency laparotomy. Dilated ileum and a hard, constricting cecal mass were found intraoperatively. She had a right hemicolectomy and ileo-transverse anastomosis, with progressive improvement postoperatively. Histology of the resected bowel segment confirmed cecal endometriosis. In conclusion, cecal endometriosis is a rare cause of intestinal obstruction. A high index of suspicion is required for diagnosis, especially if the woman is premenopausal with a history of abdominal pain that worsens with menstrual periods. Outcome is good with appropriate surgical intervention.

**KEYWORDS:** *Cecal endometriosis, cyclical abdominal pain, endometrioma, intestinal obstruction*

intestine (especially the distal ileum), and the cecum account for 70%, 1%–7%, and 3.5%, respectively.<sup>[5-7]</sup>

The precise pathogenic mechanism of endometriosis is unknown.<sup>[4]</sup> Some factors have been postulated as the cause of endometriosis.<sup>[1]</sup> Of these, two major theories were proposed. These are the metastatic and metaplastic theories. Metastatic refers to presence of endometrial tissue outside the uterine corpus. The postulated underlying mechanisms include the retrograde menstruation through the fallopian tubes may mediate spread of endometrial tissue to the peritoneal cavity, implantation of endometrial tissue following surgery as it may occur in endometriosis of the cervical mucosa, and spread of endometrial tissue through hematogenous and lymphatic routes to distant site. Metaplastic theory

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includes that the coelomic epithelium *in utero* is known to give rise to the Mullerian duct and uterus. It also forms the mesothelium of the pelvis or abdomen, and thus endometrium could arise directly from it.<sup>[1]</sup> The neurologic hypothesis is a new theory in the mechanism of development of endometriosis. From the primary lesion, the endometriotic lesion moves at a distance along the nerves to infiltrate the large bowel.<sup>[8]</sup> Other postulated factors include genetic, hormonal, and immune.<sup>[1]</sup>

This report will present our own experience with a case of cecal endometriosis with features of intestinal obstruction and with some review of literature where applicable.

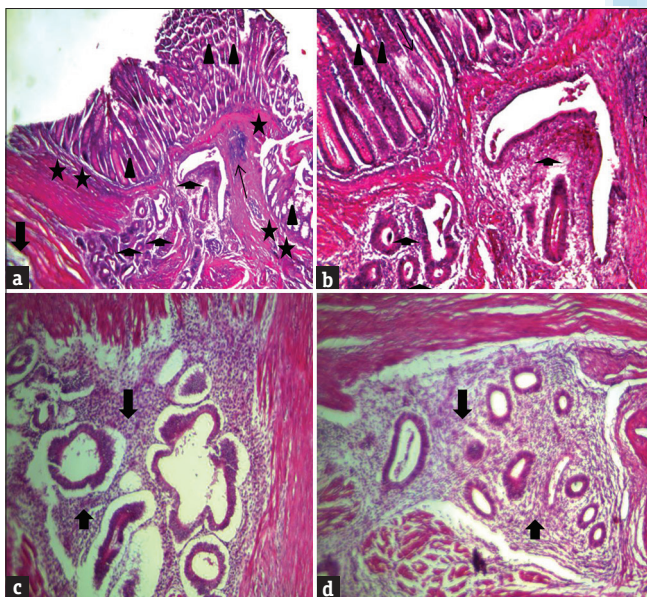
### CASE REPORT

A 42-year-old female presented with a change in bowel habits of 6 months' duration. This was associated with constipation and occasional passage of hard stool, abdominal pain, abdominal swelling, anorexia, weight loss, and easy satiety. She was passing stool at a frequency of once in 2 days as against the usual passage of stool once daily. The abdominal pain was localized to the lower abdomen, was gripping, worse during her menstrual cycle, and was relieved by analgesics. There was no history of low back pain and dyspnea. There was also no family history of cancer. She had myomectomy 2 years prior to presentation, for heavy, painful menstrual periods. Postmyomectomy, histopathology confirmed leiomyoma uteri.

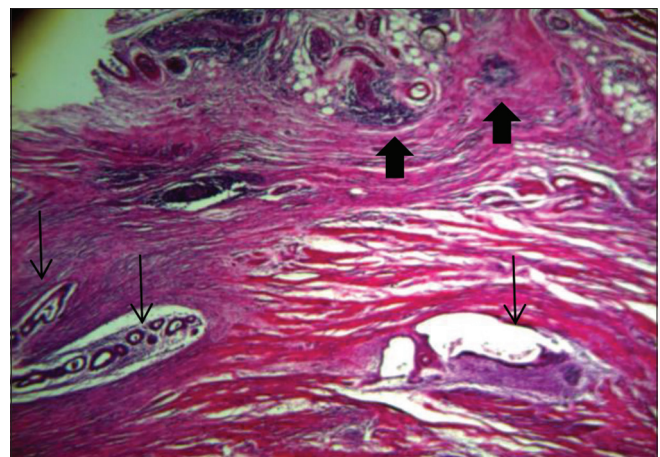
Examination revealed an acute-on-chronically ill-looking middle-aged female, afebrile, anicteric, acyanosed, not pale, and with no peripheral lymphadenopathy and pedal edema. The abdomen was full, moved with respiration, and was soft with generalized tenderness, but there was no rebound tenderness. The liver and spleen were not palpable and the kidneys were not ballotable. There were no palpable intra-abdominal masses. Ascites was not demonstrable by shifting dullness. The bowel sounds were hyperactive. The rectum contained scant fecal matter on digital rectal examination. Examinations of other systems were essentially normal. The full blood count, serum electrolyte, urea, and creatinine requested were all within normal values for her age and gender. An impression of intestinal obstruction was made. She was worked up for an emergency laparotomy as soon as possible, i.e., after resuscitation of the patient was optimal.

On surgery, the intraoperative findings were a dilated, terminal ileum and a hard, constricting cecal mass. A right hemicolectomy with ileotransverse anastomosis was done. She did well postoperatively and she was subsequently discharged home on the 10<sup>th</sup> day postoperation, after removal of stitches.

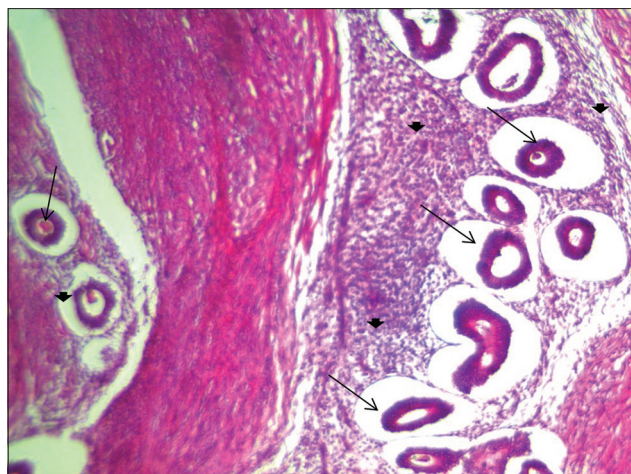
The histopathology report consists of (but is not limited to) the gross, microscopic examination findings and the final diagnosis. Grossly, the right hemicolectomy specimen consists of two resected bowel segments measuring 32 cm and 14 cm in length. The former consists of the terminal ileum, cecum, and proximal part of the ascending colon, while the latter was made up of the remaining part of the ascending colon and a portion of the transverse colon. Cut section of the former through the antimesenteric border revealed an occluded cecal lumen by a mass that is 12 cm and 14 cm from the



**Figure 1:** Histologic sections showing submucosal endometrioma (arrow heads) that has pushed the mucosa (double triangle) and submucosa (star) above the surrounding or adjacent mucosa (triangle) and submucosa (double star). Infiltrates of lymphocytes (arrow) present in the submucosa adjacent to the endometrioma and endometrial tissue (thick arrow) are seen deep into the intestinal wall. (a) (H and E, ×40), (b-d) (H and E, ×100)



**Figure 2:** Histologic sections showing endometrial glands and stroma (thin arrow) located in the muscularis propria. Infiltrates of lymphocytes are present within the muscular wall (thick arrow). (H and E, ×40)



**Figure 3:** Histologic sections showing endometrial glands (arrow) and stroma (arrow head) located in the muscularis propria. (H and E, ×100)

distal and proximal resection margins, respectively. This mass is gray, firm, and measures 6 cm × 3 cm × 3.5 cm. Cut section of the latter revealed a patent lumen.

Histologic sections showed widely distributed endometrial-type glands and stromal tissue embedded in the submucosa and muscularis propria. The overlying large intestinal-type mucosa had a bulge into the lumen in areas where endometrial tissue is located in the submucosa. Infiltrates of chronic inflammatory cells were also seen (lymphocytes). A diagnosis of cecal endometriosis was made [Figures 1-3].

## DISCUSSION

In 1860, von Rokitansky was the first to describe endometriosis.<sup>[9]</sup> Endometriosis is a benign nonneoplastic gynecological lesion seen mainly in women of reproductive age group that is characterized by the presence of functional endometrial tissue consisting of glands and/or stroma located outside the corpus uterus, in places that are physiologically inappropriate.<sup>[6,10,11]</sup>

The index case occurred in a 42-year-old woman. Usually, intestinal endometriosis is diagnosed 10 years after the development of pelvic endometriosis.<sup>[12]</sup> It, therefore, follows that the index case may have developed pelvic endometriosis when she was about 32 years of age. This age falls within the age range of 17–52 years, which has been reported for the development of endometriosis.<sup>[13]</sup> The age of the index case is comparatively similar to the higher limit (43 years) of the mean age (36 ± 7 years) of patients that develop endometriosis.<sup>[13]</sup>

The index case presented with lower abdominal pain which was aggravated during her monthly menstrual periods. Cyclical abdominal pain as observed in our own experience is consistent with the observation of most documented literatures.<sup>[5,13-17]</sup> Contrary to

this observation, De Ceglie *et al.*<sup>[6]</sup> reported in their case report of acute small bowel obstruction due to endometriosis that the abdominal pain is noncyclical, and thus bears no relationship to the menstrual periods. On presentation, there were constipation, abdominal pain, and distension with tenderness. These signs and symptoms in addition to tenesmus, diarrhea, rectal bleeding, and pain during defecation are indications or pointers to bowel endometriosis,<sup>[6]</sup> especially when the abdominal pain initially bears a relationship to the menstrual cycle.<sup>[16]</sup> Unlike, in this case, vomiting and constipation alternating with diarrhea have been reported.<sup>[6,18]</sup> This disparity may be due to the uncommon site of the lesion in the cecum in this case. Weight loss in bowel endometriosis is a variable symptom which could be attributed to the degree of obstruction and the time interval between the onset of severe symptoms and the patient's visit to the hospital.<sup>[18]</sup>

According to the European Society of Human Reproductive and Embryology guidelines, the gold standard for the diagnosis of endometriosis entails the use of laparoscopy visualization and histologic confirmation of the presence of endometrial tissue (endometrial gland and/or stroma).<sup>[19,20]</sup> The drawback of this guideline is the invasive nature of laparoscopy. Noninvasive ultrasound scan and magnetic resonance imaging (MRI) are alternatives preferred by attending clinicians.<sup>[16]</sup> The sensitivity of MRI in the diagnosis of endometriosis of the bowel is high (77%–93%).<sup>[6]</sup> Despite this, MRI is not sensitive enough for definitive diagnosis,<sup>[3]</sup> thus making laparoscopy the gold standard<sup>[7]</sup> and the only investigation prior to laparotomy that is able to confirm the presence of intestinal endometriosis.<sup>[3]</sup> Ultrasound scan (transvaginal or transrectal) shows intestinal nodules appearing as irregular, hypoechoic masses which penetrate the intestinal wall.<sup>[21]</sup> Endoscopic biopsies for histology are inadequate to make a diagnosis of endometriosis because biopsies are limited superficially to the mucosa to give nonspecific chronic mucosa changes, whereas endometriosis involves deep layers of the bowel wall.<sup>[5,12]</sup> Serum markers for the diagnosis and monitoring of endometriosis have been proposed. In monitoring the progress of endometriosis, CA-125 is more sensitive than CA-19-9, while interleukin-6 is more specific and sensitive than the former.<sup>[6]</sup> The index case presented as an acute abdomen, with little or no indications for the above investigations since intestinal endometriosis was not a consideration.

The diagnosis of endometriosis causing complete colon obstruction is very rare.<sup>[22-24]</sup> In order to make this diagnosis, a high index of suspicion should be entertained, especially if the woman is premenopausal

with a history of abdominal pain that worsens with menstrual periods.<sup>[5,7,18]</sup> Total obstruction of the gastrointestinal tract occurs in <1% of cases of endometriosis with bowel obstruction,<sup>[25]</sup> whose actual incidence is undetermined.<sup>[26]</sup> Sampson in the first quarter of the 20<sup>th</sup> century was the first to describe a case of endometriosis involving the bowel.<sup>[13,18,23,24]</sup> Endometriosis acts as if it were a malignant process, by invading contiguous or distant tissue or organ despite the fact that it is a completely benign nonneoplastic lesion.<sup>[3]</sup> Bowel endometriosis often begins as asymptomatic small serosal implant<sup>[13,17,24]</sup> that can progress to become symptomatic.<sup>[24]</sup> Endometriosis is an estrogen-dependent inflammatory disease.<sup>[24]</sup> This hormone stimulates the endometrial implants leading to cyclical bleeding. The free hemosiderin probably serves as an irritant that provokes an inflammatory reaction;<sup>[18]</sup> however, prolonged exposure to a potential irritant, be it exogenous or endogenous, can result in chronic inflammation.<sup>[27]</sup> Chronic inflammation is characterized by active inflammation, tissue destruction, and an attempt at healing.<sup>[27]</sup> Healing can be either by regeneration or fibrosis.<sup>[27]</sup> Healing occurs by fibrosis in the inflammatory reaction caused by cyclical bleeding. Fibrosis over time causes constriction of the bowel lumen in a napkin-ring fashion,<sup>[18]</sup> and intestinal obstruction may occur as a long-term complication,<sup>[17]</sup> perhaps this duration may account for cases of obstructive symptoms secondary to intestinal endometriosis seen in postmenopausal women. Menopause heralds the loss of ovarian hormonal (estrogen) stimulation with associated cessation of the cyclic hemorrhages within the gastrointestinal wall. Over time, this results in fibrosis and contraction of the bowel wall. This could lead to varying degrees of obstruction that may require resection of the stricture.<sup>[18]</sup> The histopathology showed inflammatory infiltrates, but there was, however, no significant fibrosis reported. This suggests that another mechanism may be responsible for the significant intestinal obstruction observed in the index case. Obstruction may also occur when a submucosal mass consisting of endometrial tissue (glands and/or stroma) encroaches on the bowel lumen without a true constriction of the bowel. The mass of the submucosal endometrial tissue is called an endometrioma.<sup>[18]</sup> The invasion of the bowel mucosa by an endometrioma is relatively rare. Surgery is invaluable in making the accurate diagnosis.<sup>[7]</sup> The histopathological findings in this case are consistent with an endometrioma as the cause of obstruction. Histopathology showed extensive endometrial tissue within the submucosa and muscularis propria. The latter location may not have encroached on the mucosa, reducing the lumen, but its presence within

the muscle wall may have affected the function of the smooth muscles in terms of peristaltic relaxation. It may also be that a form of resistance to the increase in the bulk of the affected bowel muscular wall may play a role.

The indication for surgical intervention includes acute abdomen secondary to intestinal obstruction, change in bowel habits, bleeding, and chronic pain.<sup>[28]</sup> The index case presented with acute abdomen following intestinal obstruction from cecal endometriosis and surgical resection was done.

## CONCLUSION

Cecal endometriosis is a rare cause of intestinal obstruction. A high index of suspicion is required for diagnosis, especially if the woman is premenopausal with a history of abdominal pain that worsens with menstrual periods. Outcome is good with appropriate surgical intervention.

## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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## Conflicts of interest

There are no conflicts of interest.

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