



Small bowel obstruction due to an endometriotic ileal stricture with associated appendiceal endometriosis: A case report and systematic review of the literature



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ABSTRACT

INTRODUCTION: Endometriosis is defined as the presence of functional ectopic endometrial tissue outside the uterine cavity. It rarely involves the small bowel and obstruction due to the same is highly uncommon. Preoperative diagnosis is difficult based on clinical and radiological studies. Diagnosis can be confirmed only on histopathological examination of the surgically resected specimen.

PRESENTATION OF CASE: A 44 years old lady presented with repeated episodes of abdominal pain, non bilious vomiting and diarrhea. She also gave history of abdominal pain during every menstruation. She had diffuse abdominal tenderness and the computed tomography showed a concentric infective/inflammatory thickening of the distal ileum. Colonoscopy confirmed a tight distal ileal stricture. After a failed trial of conservative management, she underwent a laparoscopic right hemicolectomy. The histopathological examination revealed multiple endometriotic foci in the ileum and the appendix.

DISCUSSION: Ileal endometriosis presenting as obstruction is uncommon and very few cases have been reported thus far. The symptoms are usually cyclical but may later become continuous with the progression of the disease. Preoperative diagnostic dilemma is due to the clinical and the radiological similarities to inflammatory, infective and irritable bowel diseases.

CONCLUSION: Ours is probably the first case of small bowel obstruction due to ileal and appendiceal endometriosis that was managed with laparoscopic right hemicolectomy. We highlight the preoperative diagnostic dilemma and the progression of the cyclical symptoms. Thus, endometriosis must be considered in cases of small bowel obstruction in women in the reproductive age group as a rare cause.

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1. Introduction

Endometriosis is defined as the presence of functional ectopic endometrial tissue outside the lining of the uterine cavity. It affects 4–17% of women in the reproductive age [1]. Endometriosis affecting the gastrointestinal tract has been reported in 3–37% of menstruating women. It is seen in the recto-sigmoid, caecum, small bowel and the Appendix in decreasing order of frequency. Involvement of the ileum is quite rare and it causing small bowel

obstruction is highly uncommon [2–4]. Only few case reports have been reported so far, all requiring emergency surgery. However, laparoscopic management of such disease involving a terminal ileal stricture with appendicular involvement causing subacute partial mechanical obstruction has not been reported to the best of our knowledge.

We report one such case of small bowel obstruction that was clinic-radiologically diagnosed as possible Crohn's disease but was histopathologically found to be due to endometriosis. We also highlight that cyclical abdominal pain with increasing intensity may hint towards endometriosis in a menstruating patient as a cause of small bowel obstruction.

2. Case report

A 44 year old lady presented with severe generalized abdominal pain since two days along with multiple episodes of non-projectile,

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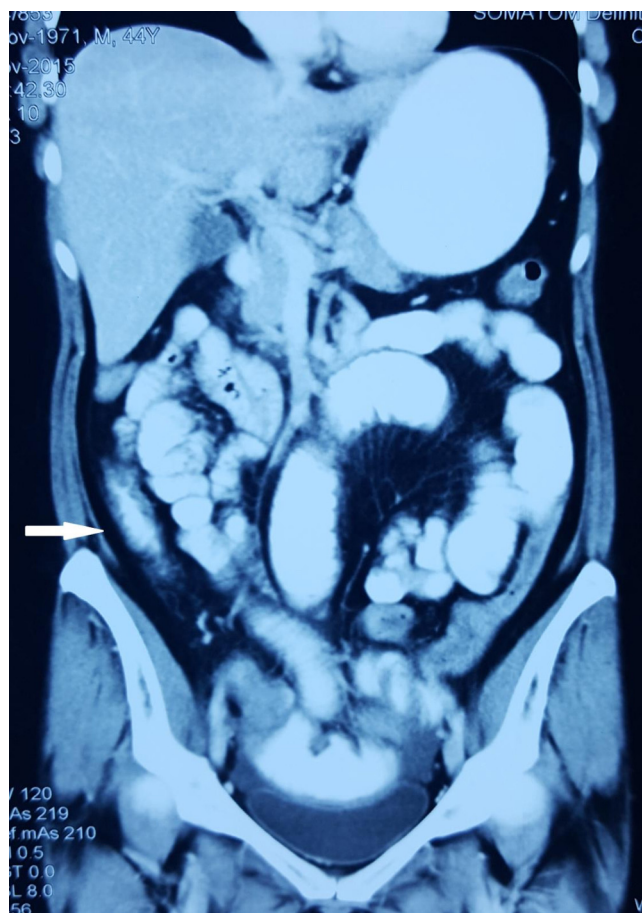


Fig. 1. CT showing concentric thickening of the distal ileum with significant luminal narrowing of the ileocaecal junction and terminal short segment of ileum with proximal ileal dilatation. No complete mechanical obstruction seen. Interbowel fluid present in pelvis.

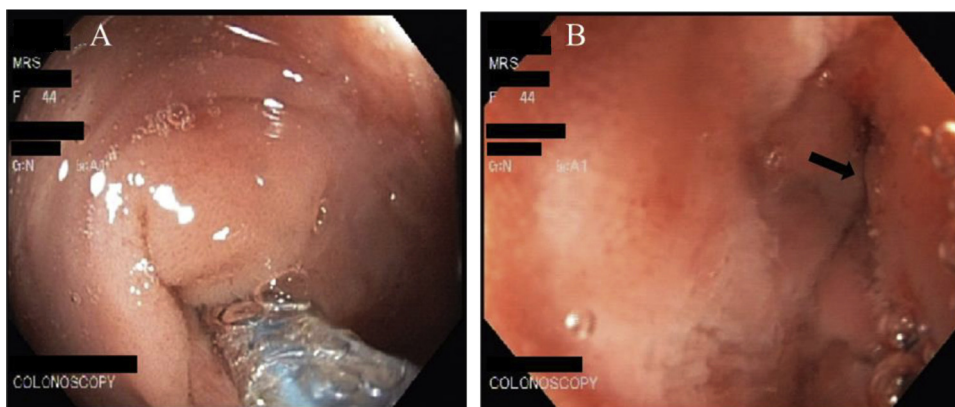


Fig. 2. A—Deflated balloon after attempted stricture dilatation, B—failure of balloon dilatation.

non-bilious vomiting and diarrhea. She had been admitted in another institution for similar complaints twice in the past month. On both occasions, she was managed conservatively for gastroenteritis with bowel rest and antibiotics. She complained of abdominal pain during every menstruation since six months that reduced with oral analgesics. However, the intensity of pain would increase with every cycle and since two months; she also experienced associated diarrhea and tenesmus. She was not on any contraceptive pills. She had undergone a caesarean section ten years ago. She was otherwise stable, her abdomen was soft with diffuse tenderness and bowel sounds were present. Her abdominal

radiograph showed small bowel obstruction with faecal loading in the ascending colon. Computed Tomography (CT) revealed a concentric infective/inflammatory thickening of the distal ileum with significant luminal narrowing of the ileocaecal junction and terminal short segment of ileum with proximal ileal dilatation. No complete mechanical obstruction was seen. Mild edematous thickening of the bowel loops with prominent vasa recta, mild surrounding fat stranding and inter bowel fluid (Fig. 1). The findings were suggestive of a subacute partial small bowel obstruction probably due to Crohn's disease. The colonoscopy showed a tight short ileal stricture just inside the ileocaecal junction. Balloon dilatation

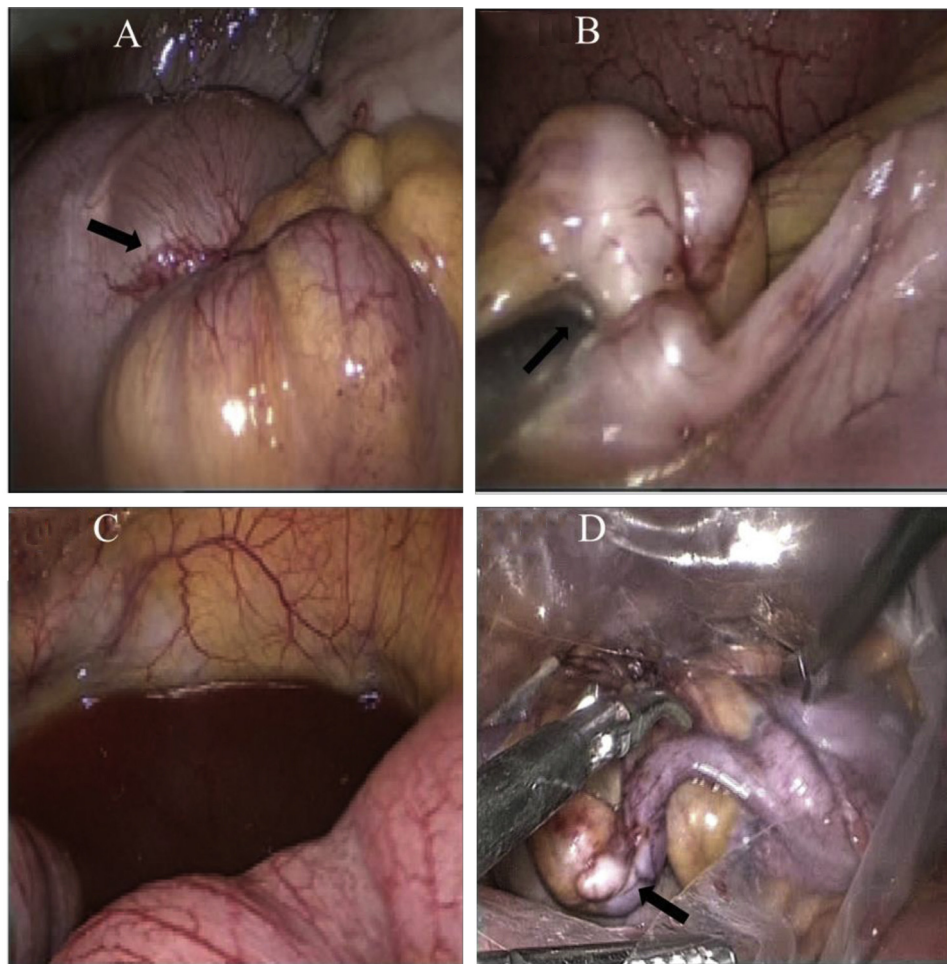


Fig. 3. A—Laparoscopic view of the ileal stricture, B—laparoscopic view of the appendicular mass at the tip of the appendix, C—hemorrhagic ascites, D—endobag with specimen (arrow pointing towards appendicular mass).

was attempted. The narrowing recurred as soon as the balloon was withdrawn, hence it failed (Fig. 2). The patient was managed conservatively but she continued to be symptomatic. Three days later, a colonoscopy was repeated and this time dilatation was done upto 15 mm. Submucosal hemorrhages were seen in the colon with ileo-caecal ulceration. A differential diagnosis of tuberculosis was also considered. Despite the dilatation, her symptoms did not subside and the patient was taken for a diagnostic laparoscopy. Intra-operatively, a distal ileal stricture with multiple enlarged lymph nodes were seen. A lump was seen at the tip of the appendix, however the base appeared normal (Fig. 3). No other abnormality was found. A laparoscopic right hemicolectomy was done. Post-operatively, the patient recuperated well and was gradually put on diet. The specimen grossly showed a dilated ileal segment, with the stricture being 2.5 cm in length. Pathologically, the ileum showed multiple endometritic foci with fibroblastic proliferation with a secondary stricture. The Appendix also showed multiple endometriotic foci. The immunohistochemistry (IHC) studies showed the glands to be positive for estrogen, progesterone receptors and vimentin. The stroma was positive for CD10 and vimentin. This confirmed the endometrial origin. No mucosal involvement was seen (Fig. 4). However, the caecum and ascending colon were free from endometriosis. Thirty lymph nodes were resected, all of which showed reactive inflammatory changes. The patient was discharged after a week on full diet. She is on regular follow-up with no cyclical complaints and is not on any hormonal therapy.

3. Discussion

The etiology of endometriosis remains unknown. It was first described in the sigmoid colon by Sampson in 1921 [5]. He popularized the theory of retrograde menstruation causing implantation and growth on the serosal surface of abdominal and pelvic organs [6]. The Minh's theory implied it to occur due to metaplastic transformation of pluripotent peritoneal mesothelium. Donnez et al., attributed it to metaplasia in the müllerian remnants [7]. Recently, a 'neurological hypothesis' implies that the lesions infiltrate the large bowel wall along the nerves distant from the primary lesion [8]. A variety of genetic alterations and unknown environmental factors contribute to the disease [9].

Endometriotic sites may be intra-peritoneal or extra-peritoneal. The intra-peritoneal sites include ovaries (30%), utero-sacral and large ligaments (18–24%), fallopian tubes (20%), pelvic peritoneum, pouch of Douglas, and gastrointestinal (GI) tract whereas the latter affect cervical portion (0.5%), vagina and recto-vaginal septum, round ligament and inguinal hernia sac (0.3–0.6%), navel (1%), abdominal scars after gynecological surgery (1.5%) and caesarian section (0.5%). Extra abdominal involvement is extremely rare [3,4,10,11]. Our patient not only had the rare ileal involvement, but also the Appendix showed endometriotic deposits. Such a dual involvement has not been reported so far.

Clinically, symptoms of endometriosis are often non specific leading to the diagnostic dilemma [12]. Gastrointestinal endometriosis present with unsettling episodes of abdominal pain,

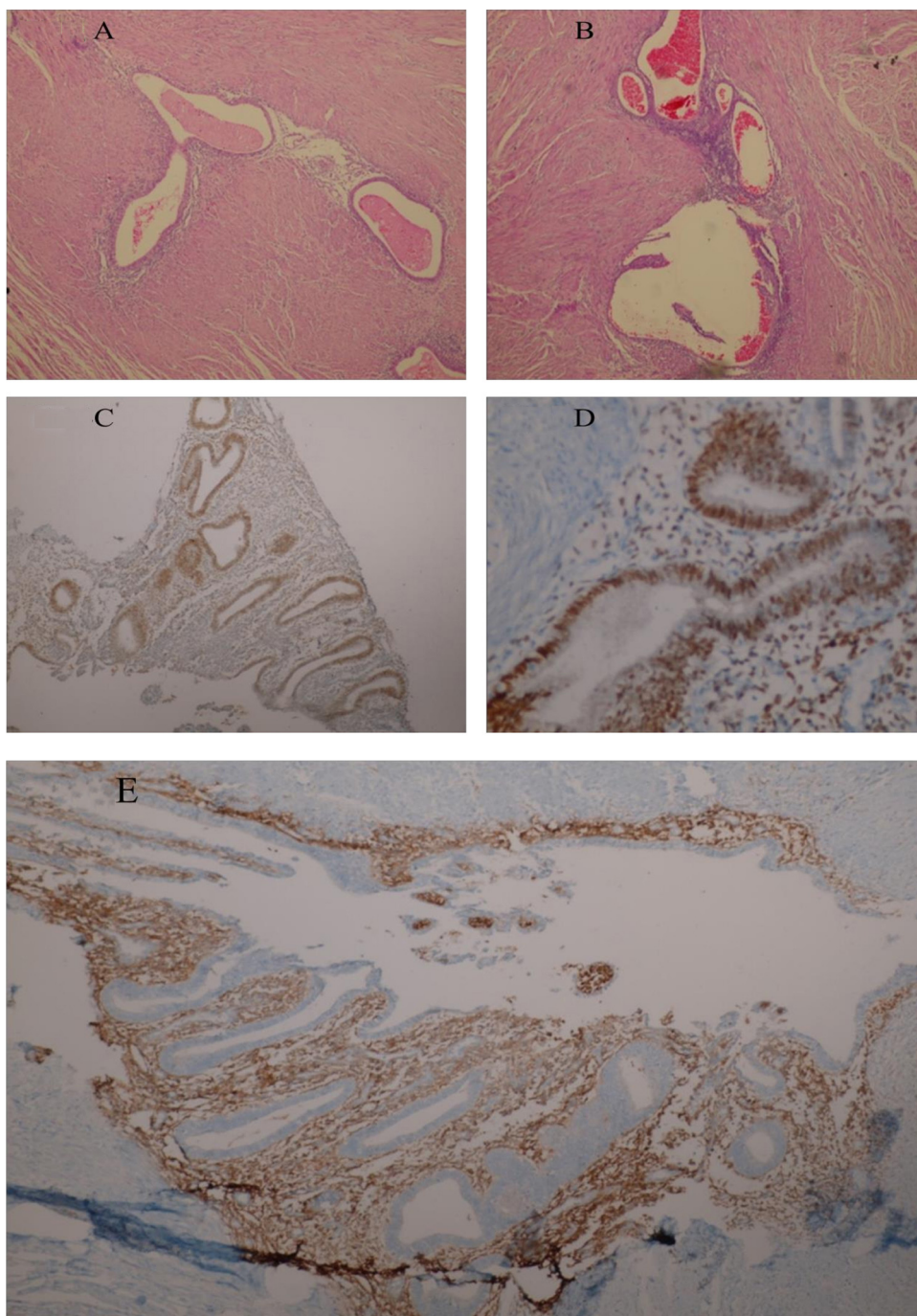


Fig. 4. A—Ileum with endometriosis, B—appendix with endometriosis, C—ER positive, D—PR positive, E—CD 10 positive.

abdominal distention, tenesmus, constipation and diarrhea [11]. The true incidence of endometriosis causing bowel obstruction is unknown [13]. Symptoms are generally cyclical but may be continuous with progression of the disease. This may point towards the rare pathology causing obstruction.

The preoperative diagnosis remains elusive due to clinical similarities to the multiple other causes of small bowel obstruction namely inflammatory bowel disease, irritable bowel syndrome, infectious and ischaemic colitis and malignancy. Intestinal endometriosis can mimic adenocarcinoma with local invasion and rapid growth [14].

Radiological imaging may be inconclusive. Since the disease does not involve the mucosa, endoscopy and barium enema is not helpful [15]. However Magnetic Resonance Imaging seems to have a

higher sensitivity [16,17]. Laparoscopy is considered gold standard. The diagnosis can be confirmed only on histology.

Small bowel endometriosis is seen only in bowel serosa and deposits are usually not larger than 2 cm in size. The mucosa is not involved probably due to poor nerve supply. Patchy 'grey glistening' lesions are seen [17]. Lymph nodal involvement has also been reported attributing to lymphatic dissemination from the intestinal endometriotic foci [18,19]

Gastrointestinal endometriosis is usually found as an incidental finding on laparotomy. Asymptomatic and mildly symptomatic cases may be treated using hormonal treatment. However, acute obstructive cases may be dealt with surgery [11]. Suspicion of malignancy may also warrant a radical resection.

Table 1
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S. No.	Author & year	History & clinical findings	Differential diagnosis	Diagnostic modalities & findings	Treatment	Outcome
1.	Alistair A.P. Slessor, 2010	Abdominal pain with constipation since 1mth. 3LSCS, on OCP,	Crohn's disease, Tuberculosis	CT—small bowel obstruction with a transition point at terminal ileum	Right hemicolectomy	Uneventful
2.	Kerem Karaman, 2012	27yrs female with abdominal pain and diarrhea since 3 months	Crohn's disease	Colonoscopy: Inflammatory changes CT: 2 cm lesion in ovary with dilated small bowel loops	Conservative treatment for Crohn's, symptoms persisted. Exploratory laparotomy with resection of involved ileum and end ileostomy	Uneventful
3.	Alatise O.L., 2010	34 yrs female with abdominal pain, incisional hernia and suprapubic mass. Also had secondary infertility and had undergone myomectomy.	Adhesive intestinal obstruction	CT: incisional hernia with imminent adhesive small bowel obstruction.	Exploratory laparotomy with small bowel resection	Uneventful
4.	Cappell M.S., 1991	38 years female with abdominal pain.	Crohn's disease	CT: suggestive of Crohn's ileitis	Exploratory laparotomy with resection of the involved small bowel	Uneventful
5.	Kavitha Duraisamy Yogini, 2015	37years female with primary infertility with abdominal pain since 1 year, increased since 1 week. No cyclical pain	Crohn's disease	Colonoscopy: patchy infiltration throughout colon, biopsy suggestive of nonspecific colitis. Diagnostic laparoscopy done 3 months back showed ileal strictures. CT: adhesive small bowel obstruction	Laparoscopic ileal resection	Uneventful
6.	Haluk Recai Unalp, 2012	45 years female with abdominal pain, vomiting	Acute abdomen	USG: thickened, aperistaltic and distended loops of the small bowel.	Laparotomy: right hemicolectomy for a tumoral mass 5 cm proximal to ileo-caecal junction. Left salpingectomy for hydrosalpinx	Uneventful
7.	Jeffrey R. Ridha, 2003	36 years female with abdominal pain and vomiting since 1 day	Acute ileal obstruction	Abdominal radiograph: air fluid levels. CT: massively dilated small bowel loops	Exploratory laparotomy: ileal obstruction with chocolate cysts in b/l ovaries, endometrial tissue on sigmoid and appendix. Small bowel resection with b/l ovarian cystectomies, appendectomy	Uneventful
9.	Collins, 2016	40 years lady with dysmenorrhoea and lower abdominal pain.	Regional ileitis and carcinoid of ileum	–	distal ileum inflamed and twisted on itself.	Uneventful
10.	Soumekh A., 2015	44 years lady with abdominal pain and constipation. No dysmenorrhoea	Partial small bowel obstruction	CT: small bowel obstruction proximal to ileo caecal junction	Resection and anastomosis	Uneventful
11.	Attar A., 2007	Symptoms of intestinal obstruction	Intestinal obstruction	CT: ileal obstruction	Ileo colonic resection and anastomosis	Uneventful
12.	Arachchi A., 2011	39 years lady with cyclical right iliac fossa pain	Adhesive intestinal obstruction	Abdominal skiagram: fecal loading in ascending colon. CT: mid to distal ileal dilatation, no transition point.	Stenosed and fibrosed terminal ileum and caecum. Adhesiolysis with resection and anastomosis.	Uneventful.
13.	Thanassery R.B., 2008	43 years lady with recurrent periumbilical pain, bilious vomiting and constipation	Small bowel obstruction	CT: short segment ileal stricture	Resection of the involved ileum with end ileostomy	Uneventful

The commonest site of gastrointestinal endometriosis is the rectosigmoid. Isolated involvement of the ileum has also been reported in 1–7%. The disease causing obstruction is rare, less than 1% [20]. Dual involvement of the terminal ileum and the Appendix

is extremely rare. The incidence of intestinal resection for bowel obstruction is 0.7% [20]. Very few reports describe laparoscopic management of the disease causing obstruction.

The management includes hormonal therapy and surgery. The former with danazol or gonadotrophin-releasing hormone analogs may be used in patients without obstruction. However, resection of the involved bowel remains the choice of treatment for complicated or unresolved cases.

Table 1 summarizes similar cases that have been reported so far. Most cases presented with either partial or complete small bowel obstruction attributed to other causes. Most underwent emergent surgery. Hormonal therapy is not popular in acute cases of obstruction.

Our case highlights the progressive nature of the disease. Our patient earlier had pain only during menstruation which later progressed to repeated episodes of crampy pain with diarrhea and tenesmus. Despite, endoscopic balloon dilatation of the ileal stricture, the symptoms continued. The tight terminal ileal stricture was eventually managed by laparoscopic right hemicolectomy. Histology confirmed the diagnosis.

4. Conclusion

Endometriosis as a cause of small bowel obstruction with appendicular involvement remains a rare entity. A preoperative diagnosis is difficult due to lack of pathognomic clinical and radiological findings. However, a high index of suspicion is required in women in the reproductive age group. Cyclical symptoms of increasing intensity may hint towards the diagnosis.

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