

Intestinal Obstruction due to Rectosigmoid Endometriosis Mimicking Stenotic Rectosigmoid Cancer: A Case Report

*Agbonrofo PI,¹ Brotobor O,¹ Udoh MO,² Odigie VI¹

ABSTRACT

Endometriosis is the presence of endometrial-like tissues (glands and stroma) outside the uterine cavity. It is a common cause of morbidity in women of reproductive age (up to 10%). Endometriosis may affect a variety of organs including the intestines (5-12% of endometriotic women). About 90% of intestinal endometriosis involves the rectosigmoid colon and may be symptomatic or asymptomatic. Bowel obstruction occurs in less than 1% of patients with endometriosis. This is a report of a 32-year-old female who presented with altered bowel habits of 6 months, abdominal pain with obstipation of 11 days, vomiting of 10 days, and abdominal distension of 8 days. She has had severe cyclical lower abdominal pains for two years and primary infertility of 6 years. Physical examination findings were in keeping with intestinal obstruction. She was optimized for surgery with intraoperative findings of a hard complex mass comprised of the rectosigmoid colon, uterus and adnexae; dilated proximal colon and collapsed Small intestines. She had a multi-stage resection and anastomosis of the rectosigmoid colon and bilateral wedge resection of the ovaries. Histology of the resected specimen revealed endometriosis. She is doing well after discharge with relief of symptoms; however, she is still being managed for Infertility. In conclusion, endometriosis though benign may have crippling physical and/or psychosocial effects on women of reproductive age. Recto-sigmoid endometriosis may be considered a rare differential diagnosis of bowel obstruction from recto-sigmoid cancer and may require a multi-stage surgical approach as appropriate. Management of endometriosis-associated infertility is challenging.

Keywords: *Intestinal Endometriosis, Endometriosis, Intestinal Obstruction, Recto-sigmoid Cancer, Rectosigmoid Endometriosis, Infertility*

INTRODUCTION

Endometriosis is the presence of endometrial-like tissues (glands and stroma) outside the uterine cavity.¹ It is a common cause of morbidity in women, particularly in their reproductive period. It is present in up to 10% of women of childbearing age.¹ It was thought to be a rare occurrence in African women in the past but its diagnosis is becoming more common with time.² This disease, though benign, may be a cause of serious complaints such as chronic pelvic pain, dysmenorrhea, dyspareunia and Infertility.

About 40% of women with infertility have associated endometriosis.^{3,4} Endometriosis may affect a variety of organs

including the ovary (the commonest site of endometriosis), fallopian tubes, peritoneum, ureter, urinary bladder, intestines, etc. Intestinal involvement is present in about 5% to 12% of endometriotic women.^{5,6} About 90% of intestinal endometriosis involves the rectosigmoid colon.^{5,6} Intestinal endometriosis, just like endometriosis elsewhere, may be symptomatic or asymptomatic.

Symptomatic intestinal endometriosis may present with non-specific (dysmenorrhea and dyspareunia) or specific symptoms. The bowel-specific symptoms depend on the site of the lesion, how large the lesion is and the depth of intestinal wall involvement. The specific symptoms related to the presence of the lesion in the rectosigmoid colon are mainly abdominal pain and changes in bowel habits (diarrhoea, constipation, dyschezia, tenesmus, bleeding per rectum and intestinal obstruction).¹ Bowel obstruction occurs in less

Department of Surgery,¹ University of Benin Teaching Hospital, Benin City, Nigeria.

Department of Pathology,² University of Benin Teaching Hospital, Benin City, Nigeria.

*Corresponding author: pagbonrofo@gmail.com, peter.agbonrofo@uniben.edu.

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than 1% of patients with endometriosis.⁷ These bowel-specific symptoms may be present in other bowel pathologies such as irritable bowel syndrome, rectosigmoid cancers, Crohn's disease, diverticular disease, adhesive bowel disease, solitary rectal ulcer, etc.

Preoperative diagnosis of intestinal endometriosis is more often than not a diagnosis of exclusion, especially in patients with no prior diagnosis of Endometriosis. A history of cyclical symptoms of dysmenorrhea, dyspareunia and pelvic pain may be suggestive of a diagnosis of Endometriosis.¹ Confirmatory diagnosis of intestinal endometriosis is the presence of endometrial tissue in the biopsy specimen.²

CASE PRESENTATION

She is a 32-year-old lady, a primary school headmistress, nulliparous first seen at the Emergency room with altered bowel habit (increasing constipation) of 6 months, abdominal pain with obstipation of 11 days, vomiting of 10 days, abdominal distension (gradual onset and generalised) of 8 days. She has had severe recurrent lower abdominal pains during menstruation (dysmenorrhea) for two years and has been married for 6 years with primary infertility.

She was febrile, dehydrated, tachypnoeic and tachycardic. Her abdomen was generally distended, tensed and tender. The rectum contained mucus.

She was optimised for surgery and had an emergency laparotomy with transverse loop colostomy. The intraoperative findings included; hard complex mass comprised of the rectosigmoid colon, uterus and adnexae; dilated proximal colon and collapsed small intestines. She had good postoperative recovery and uneventful postoperative management and was discharged on the 17th postoperative day.

A double-contrast barium enema (10th postoperative day) showed retrograde filling outlining the distal rectosigmoid colon with a

complete holdup in the lower sigmoid colon where there was an irregular filling defect. Colonoscopy (14th postoperative day) showed sharp cut-off/ obstruction about 25cm from the anal verge. Rectal mucosa was normal. The distal limb of the loop colostomy showed cut off in the lower sigmoid; no mass, no lesion and no ulceration.

Transvaginal ultrasound scan (3 months post-operation) showed non-gravid anteverted Uterus measuring 6.2cm x 4.0cm x 3.9cm with uniform myometrium and a demonstrable mid-line echo; there were multiple varying-sized complex right adnexa masses, one measuring 2.8cm x 3.3cm x 3.7cm with a volume of 17.7mls containing multiple internal septations with another measuring 3.2cm x 2.3cm x 3.3cm with a volume of 12.6mls containing echogenic nodule. The left adnexa was unremarkable.

Three months after the initial surgery, she had a laparotomy with resection of the rectosigmoid colon (with colorectal anastomosis) and bilateral wedge resection of the ovaries. The intraoperative findings included bilateral tubo-ovarian masses; multicystic ovaries with the right ovary measuring 8cm x 6cm and the left 6cm x 4cm; bilateral hydrosalpinges. There was also a hard, structured rectosigmoid colon. The postoperative management was satisfactory and she was discharged home on the 11th postoperative day. Histologic examination of the excised rectosigmoid colon and ovaries revealed endometriosis. (Figures 1, 2 & 3).

The transverse loop colostomy was reversed five months after the first surgery. She was discharged home on the 11th postoperative day. She has had several clinic visits with complete relief of bowel symptoms and markedly reduced dysmenorrhea. She desires to achieve pregnancy but yet to conceive and is still being managed by the gynaecologist for infertility.

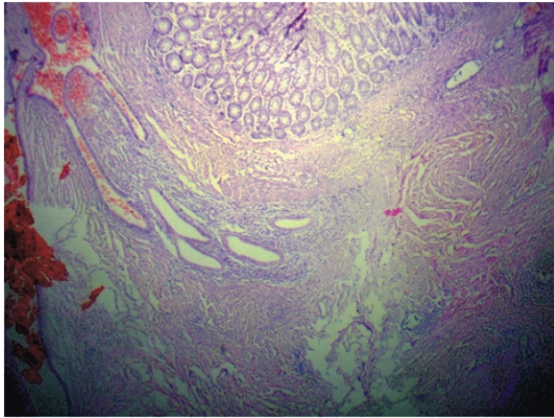


Figure 1: Photomicrograph of rectosigmoid tissue sections (H&E x 40) showing endometrial glands and stroma, with haemorrhage in the submucosa (centre and left), overlying normal intestinal mucosa glands in top centre field.

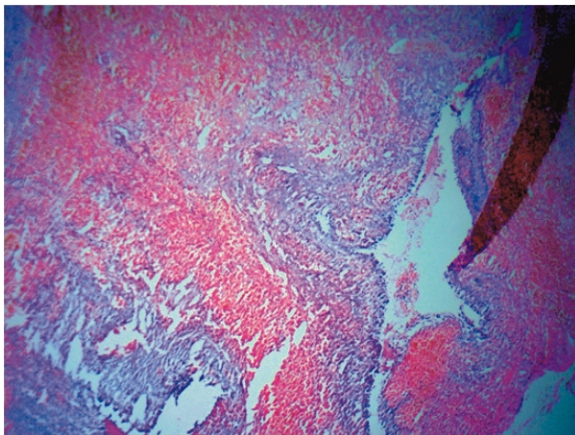


Figure 2: Photomicrograph ovarian stroma (H&E x 100). Haemorrhage and cystic endometrial type gland within ovarian stroma.

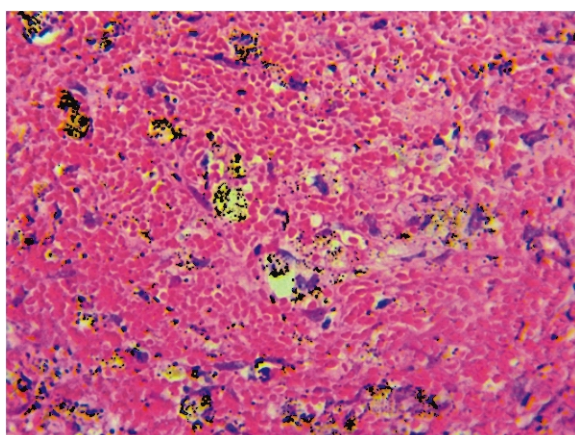


Figure 3: Photomicrograph of ovarian stroma (H&E x 400). Haemorrhagicnecrotic focus showing numerous hemosiderin-laden macrophages indicating chronic bleeding.

DISCUSSION

Endometriosis runs a chronic course with potentially debilitating effects on the well-being of the affected patient (both physical and psychosocial) during their reproductive and productive (economic) life period. A lot of resources are expended on the care for these patients more so in a "pay-out-of-individual-pocket" setting as ours. The psychological impact of the disease from its chronicity, potential crippling pain and infertility may not be quantifiable in economic terms.

Bowel Endometriosis usually does not occur in isolation. It occurs in the setting of disease involving other pelvic structures (e.g. vagina wall, uterosacral ligaments with or without involvement of the torus uterinum).¹ In the index case, there was ovarian and bowel involvement. Early in the disease process, the main symptoms are deep dyspareunia and dysmenorrhea. The worsening disease may present with worsening bowel symptoms with bowel obstruction at the extreme.¹ This was present in the index case.

Preoperative diagnosis of bowel endometriosis is the exception rather than the rule. Suggestive features include cyclical dyspareunia, dysmenorrhea, fertility challenges and bowel symptoms (e.g. altered bowel habits including abdominal pains, dyschezia and rarely haematochezia (there may also be cyclical worsening of these features)).^{2,4} Physical examination including bimanual pelvic examination may be unremarkable for endometriosis even in severe disease.¹ Findings suggestive of endometriosis include a fixed retroverted uterus, tender fibrotic nodule in the adnexae (parametrium, uterosacral ligaments, vesicovaginal septum, torus uterinum, rectovaginal septum).⁸

Investigations important for the preoperative diagnosis of intestinal endometriosis include imaging studies (e.g. transvaginal ultrasonographic scan, magnetic resonance imaging, doublecontrast barium enema) and laparoscopy (to visualise the lesions, take a biopsy or even perform

definitive surgery).^{1,8} In the emergency setting of intestinal obstruction, extensive preoperative evaluation may not be feasible and expedient operative management is done (laparotomy or laparoscopy) as occurred in this case.

Definitive diagnosis of endometriosis is by histological examination of tissue obtained by invasive surgical or laparoscopic procedures.^{2,9} Endometriosis typically appears as superficial or variably deep lesions extending >5mm beneath the peritoneal or serosal surface of affected organs. They appear as black, dark-brown, or bluish-puckered lesions, nodules or cysts containing old haemorrhage, with variable extent of surrounding fibrosis. Histologically, endometriotic foci contain endometrial glands and stroma or rarely endometrial stroma alone.^{10,11} Sometimes, stromal component is obscured by foamy and pigmented histiocytes, fibrosis, elastosis, smooth muscle metaplasia, myxoid change, and decidual change, making histological diagnosis difficult, especially in long-standing cases.¹¹

Intestinal endometriosis most commonly affects the rectosigmoid colon, appendix, caecum, and distal ileum.^{5,6,10} Serosal implants may erode through the sub-serosal layers and cause marked thickening and fibrosis of the muscularis propria, but an intact overlying mucosa is almost always present.¹⁰ Extensive disease, with organizing haemorrhage may cause fibrous adhesions between tubes, ovaries, and the intestine, as seen in our patient.¹⁰

Malignant transformation is a rare complication of endometriosis, occurring in less than 1% of cases.¹⁰ Majority (approximately 75%) of these arise from endometriosis of the ovary, less common sites being the rectovaginal septum, rectum, and sigmoid colon, amongst others. Endometrioid carcinoma followed by clear cell carcinoma is the most common type.¹⁰ A rare case of endometrial stromal sarcoma in a patient with extrauterine endometriosis has been reported.¹²

There is no unifying theory regarding the origin of endometriosis.^{9,13} The fact that the pathogenetic mechanisms underlying endometriosis are yet to be defined with certainty, is the main reason why to date, there is no definitive diagnostic and therapeutic path for this disease.⁹ The most accepted theories include retrograde menstruation, haematogenous or lymphatic spread, coelomic metaplasia and extrauterine-sourced stem cells, and recently embryogenetic theory with Müllerian rest.^{9,13} Molecular hallmarks of this disease are supported collectively by investigations into the pathophysiology of endometriosis, including genetic predisposition, estrogen dependence, progesterone resistance and inflammation.¹³ These remain areas of active research with the promise of the development of novel non-surgical diagnosis and therapy.

A multidisciplinary approach to the management of these patients is important and this includes the gynaecologist, general surgeon, stoma therapist, radiologist, pathologist, etc.^{1,2,4,8} Operative management of obstructed rectosigmoid endometriosis is dependent on the preoperative clinical condition of the patient and the extent of the disease.⁸ A single-stage procedure/operation or multiple-stage approach may be used in the management of the patient. The index case had staged operations (as she presented with complete obstruction, was quite ill and had a complex inflammatory mass; transverse loop colostomy (first stage); rectosigmoid resection with colorectal anastomosis and wedge resections of the ovaries (second stage); reversal of the transverse loop colostomy (third stage). Less extensive operative procedures are done for less extensive bowel endometriosis: these include shaving of the lesion and disc excision of the lesion.⁶ Adjunct medical therapy using hormonal agents (e.g. combined oral contraceptive pills, progestins, estrogens, GnRH agonists) are helpful in pain control (dysmenorrhea, dyspareunia) and reducing disease recurrence.^{1,6}

Endometriosis reduces chances of achieving pregnancy (as seen in the index

patient with primary infertility) and also worsens pregnancy outcomes (higher risk of placental praevia, placenta abruption, hypertension, preterm birth, postpartum haemorrhage, caesarean section and postpartum hysterectomy) in those who become pregnant.¹⁴ Achieving pregnancy in the patient with endometriosis-associated infertility is challenging and this may be improved with assisted reproductive technique.⁸

CONCLUSION

Endometriosis is a common chronic disease affecting women during their reproductive lifetime. It may have a crippling effect on a woman's life both physically and psychosocially. Intestinal endometriosis, though less common, may cause bowel obstruction and may be included in its differential diagnosis, especially in women with a history suggestive of endometriosis. A multidisciplinary approach to care for patients with intestinal endometriosis is of utmost importance. After relieving bowel obstruction in those with obstructive intestinal endometriosis, care for the patient about control of chronic pain and achieving pregnancy (according to patient's wish) is paramount.

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