



Cecal endometriosis presenting as a term intrauterine fetal demise and gastrointestinal hemorrhage: A case report

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ABSTRACT

Background: Of women diagnosed with endometriosis, 3.8–37% have bowel endometriosis. The cecum is the least common site for endometriotic implants affecting the bowel, accounting for only 3.6–6% of cases. We present a case of intrauterine fetal demise at term in which the patient was found to have gastrointestinal bleeding caused by endometriosis of the cecum.

Case: A 35-year-old woman, gravida 4, para 1, at 37 weeks and 3 days of gestation, without a known history of endometriosis but with two prior miscarriages, presented with severe anemia and intrauterine fetal demise. During delivery, melanic stool was noted. Colonoscopic biopsy noted the source of bleeding to be a 2 cm endometriotic implant in the patient's cecum. Suppression therapy was started. Postpartum, the patient underwent laparoscopic cecectomy and pathology confirmed the diagnosis of endometriosis.

Conclusion: Hemorrhage from endometriotic implants may occur during pregnancy due to changes in the hormonal milieu. Bowel endometriosis may increase the risk of maternal hemorrhage during pregnancy, thereby increasing the risk of unfavorable pregnancy outcomes, including intrauterine fetal demise.

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1. Introduction

Endometriosis, the presence of viable, estrogen-sensitive, endometrial-like glands and stroma outside the uterus, affects 6–10% of reproductive-aged women [1]. Endometriosis affecting the bowel is a likely underdiagnosed condition, often only found incidentally during intra-abdominal surgery performed for a separate indication. The estimated incidence of bowel endometriosis among women diagnosed with endometriosis ranges from 3.8% to 37%, reflecting the difficulty of diagnosing bowel endometriosis. Women with bowel endometriosis are often diagnosed with other disorders; the broad differential includes irritable bowel syndrome, inflammatory or ischemic colitis, diverticulitis, malignancy, and pelvic inflammatory disease [2]. The most common site of implantation in bowel endometriosis is the rectosigmoid colon or rectum, in 70–95% of cases, followed by the small intestine, most commonly the ileum, in 7–12% of cases, and the appendix in 6–8% of cases. The least common site of endometriotic implantation is the cecum, in 3.6–6% of cases [2–4].

The pathophysiological changes of eutopic and ectopic endometrium during pregnancy are an evolving area of research. Several studies have investigated the relationship between clinically diagnosed endometriosis and adverse pregnancy outcomes, including maternal and

fetal complications [5–10]. However, the relationship between bowel endometriosis and obstetrical outcomes is less clear [11,12]. We found only two cases in the literature describing bowel endometriosis during pregnancy presenting as gastrointestinal hemorrhage [13,14]. In this report, we describe a case of endometriotic implants in the cecum presenting as intra-uterine fetal demise and gastrointestinal hemorrhage.

2. Case Presentation

A 35-year-old woman, gravida 4, para 1, without a known diagnosis or family history of endometriosis, but with two prior miscarriages, and no prior surgeries presented at 37 weeks and 3 days of gestation with nausea, tachypnea, and diarrhea for two days. The patient had a previously uncomplicated prenatal course, except for a treated *Streptococcus agalactiae* urinary tract infection during the first trimester. Her routine prenatal labs at 28 weeks of gestation were unremarkable, with normal glucose challenge testing and a hemoglobin concentration of 11 g/dL. The patient reported feeling excessive fetal movement in the morning followed by abdominal pain with four episodes of diarrhea, including some blood. The patient then began to feel weak, dizzy, short of breath, and no fetal movement. At initial presentation, intrauterine fetal demise was diagnosed. The patient had an ECG showing sinus tachycardia with a rate of 155 bpm, a negative chest X-ray, a low hemoglobin concentration of 6.1 g/dL, and an elevated white blood cell count, troponin, and lactate. Computed tomography angiogram of the chest was negative

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for pulmonary embolism. Blood pressures and oxygen saturations remained within normal limits. Influenza and respiratory syncytial virus testing were negative. The patient was started on IV piperacillin-tazobactam for suspected sepsis and was admitted for induction of labor. Given presumptive chorioamnionitis, her antibiotics were changed to ampicillin, gentamicin, and clindamycin, and her induction was started with a Cook catheter and continued with a Pitocin infusion. The patient was transfused 2 units of packed red blood cells, was continued on IV fluids, and was given an epidural for pain management. While pushing, thick melanotic stool appearing from the rectum was seen and testing confirmed the presence of blood in the stool. The patient underwent a spontaneous vaginal delivery with second-degree laceration. The fetus delivered with the cord loosely wrapped around its trunk once, but with no other obvious abnormalities.

Given the rectal bleeding noted at delivery, the gastroenterology service was consulted. Diagnostic esophagogastroduodenoscopy found no upper gastrointestinal bleeding. Colonoscopy suggested a 2 cm inflammatory mass at the cecum near the appendiceal orifice and revealed a large amount of blood-stained fluid in the colon with no active source of bleeding (Fig. 1). Biopsies taken at this time revealed granulation tissue with fibrinopurulent debris and mixed inflammation, and the site was coagulated with resolution of bleeding. Subsequent computed tomography of the patient's abdomen confirmed the presence of a 2 cm inflammatory mass, with no other abnormal findings (Fig. 2). Capsule study showed no other abnormalities. Throughout her hospitalization, a total of 6 units of packed red blood cells was transfused. Platelets and coagulation studies remained normal, as did liver function tests. Troponin peaked at 0.36 ng/mL and trended downward. Echocardiogram was normal with no wall motion abnormalities. Stool ova and parasites and stool toxin assays were negative for *Shigella* species and hemorrhagic *Escherichia coli*.

Placental cultures grew *Corynebacterium* species and *Bacteroides vulgatus*. The patient remained afebrile with no further rectal bleeding and was discharged. Chromosomal analysis was normal. No findings of chorioamnionitis were identified on fetal autopsy, suggesting the placental culture showed contamination.

Six weeks postpartum, a repeat colonoscopy again demonstrated the same 2 cm mass with biopsy suggestive of endometriosis (Fig. 3). Suppression therapy with norethindrone 0.35 mg daily was initiated. The patient underwent a laparoscopic cecectomy with primary anastomosis to remove the entire mass. Intraoperative

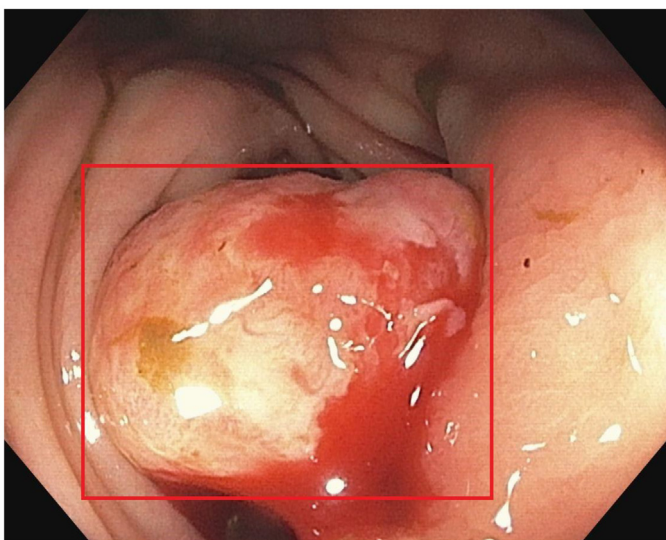


Fig. 1. Initial colonoscopy demonstrating the 2 cm inflammatory cecal mass with hemorrhage.

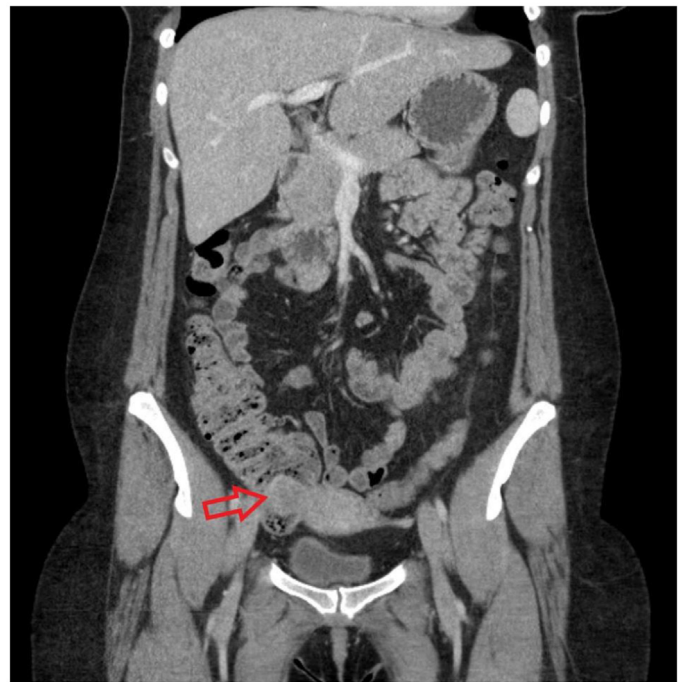


Fig. 2. Computed tomography scan demonstrating a cecal mass without other abnormalities.

inspection revealed no abnormalities of the pelvic organs (Fig. 4) or of the upper abdomen other than the cecum (Fig. 5). There were no obvious endometriotic implants; random peritoneal biopsies were taken for sampling.

Final pathology of the specimen confirmed endometriosis involving the colonic mucosa with ulcerated granulation tissue. Random biopsy of the right posterior cul-de-sac also returned endometriosis on pathology.

3. Discussion

Bowel endometriosis affecting obstetrical outcomes is a relatively under-investigated phenomenon. We believe this case is the first to describe hemorrhage from endometriotic implants in the cecum during

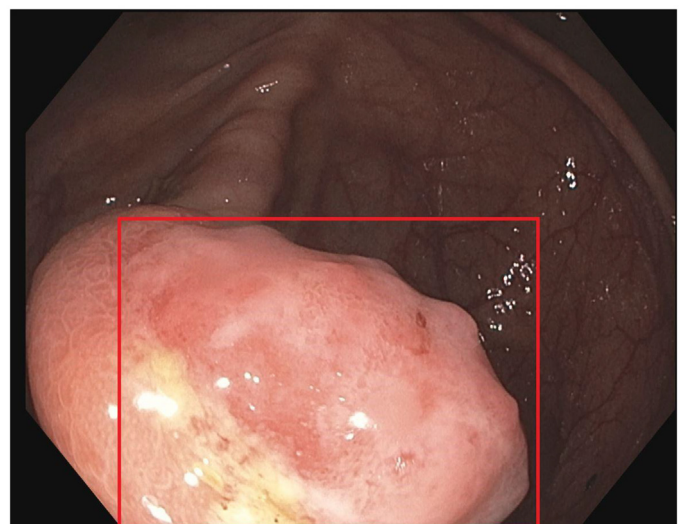


Fig. 3. Repeat colonoscopy at six weeks postpartum demonstrating the same cecal mass.

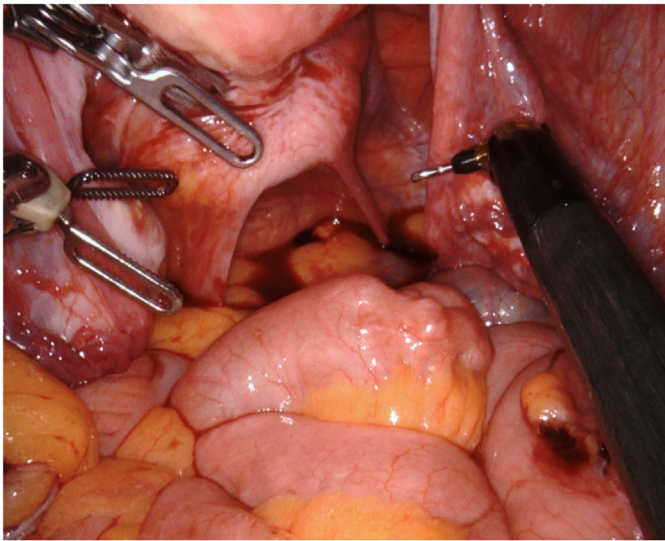


Fig. 4. Diagnostic laparoscopy demonstrating no abnormalities of the pelvic organs.

pregnancy. This case also draws attention to gastrointestinal hemorrhage from endometriotic implants that can cause adverse pregnancy outcomes, including intrauterine fetal demise.

In the presented case, bowel endometriosis and hemorrhage resulting in intrauterine fetal demise likely occurred due to maternal hemorrhage resulting in reduced uteroplacental perfusion and fetal hypoxia. Bowel endometriosis resulting in gastrointestinal hemorrhage during pregnancy provides a logical link between a reduction in maternal oxygen carrying capacity, decreased fetal perfusion, and intrauterine fetal demise. Studies have shown that severe anemia in pregnancy increases the risks of preterm birth, growth restriction, stillbirth, and neonatal death [15]. The hemorrhage sustained by the patient in the current presentation likely persisted long enough to lead to uteroplacental insufficiency. This presents the most likely acute event leading to intrauterine fetal demise for our patient.

Hemorrhage of endometriotic implants during pregnancy has been described in the literature [6,7,11,13,14]. The proposed pathogenesis of hemorrhage arising from endometriotic implants is the decidualization of implants as a response to the pregnancy hormone milieu. Implants can increase in size by almost 20% during pregnancy and become highly vascular, causing hemorrhage [6]. Many of the case reports in

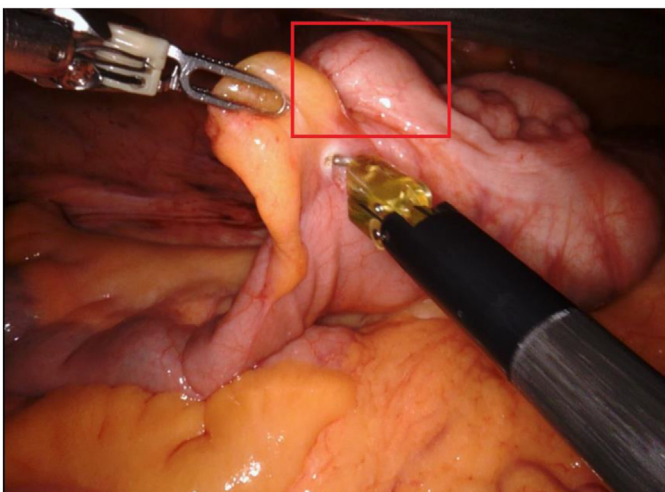


Fig. 5. Diagnostic laparoscopy demonstrating the cecal mass.

the literature describe hemorrhage secondary to endometriotic implants occurring in the third trimester [6,7,13,14]. This may suggest a causal relationship between hormonal milieu and hemorrhage from endometriotic implants.

Women with endometriosis may also have abnormally functioning endometrium and placentae, which may lower the fetal tolerance to an insult such as acute blood loss. Brosens et al. suggest that the pre-pregnancy uterine and hormonal milieu in women with endometriosis is a risk factor for placental bed disorders [6,16]. A study by Bromer et al. demonstrated that women with endometriosis achieve a significantly lower peak endometrial thickness, an observation similarly found in women with polycystic ovarian syndrome or recurrent pregnancy loss [17]. Studies suggest that “progesterone resistance” impacting the stroma, vascular smooth muscle, and endothelial cells alters the process of deep placentation, which may explain the relationship of endometriosis to implantation failure [18–20]. The typically progesterone-dependent decidualization of endometrial spiral artery segments is also abnormal in the hormonal milieu of a woman with endometriosis [6,16].

A review of endometriosis during pregnancy performed by Maggiore et al. found sixteen cases of intestinal perforation due to ectopic endometrium related to pregnancy. Of the thirteen cases of intestinal perforation during pregnancy, none were due to lesions in the cecum and only one presented with melena [7]. Thus, our report of spontaneous hemorrhage of an endometriotic implant in the cecum during pregnancy, presenting with melena and fetal demise, presents a clinically useful addition to the literature regarding bowel endometriosis and adverse pregnancy outcomes.

4. Conclusion

Bowel endometriosis in pregnancy increases the risk of gastrointestinal hemorrhage, compounding the risk of an already abnormally functioning ectopic endometrium and placenta. The further investigation of the relationship between bowel endometriosis and intrauterine fetal demise is warranted, as bowel endometriosis during pregnancy is a potentially serious risk factor for intrauterine fetal demise.

Contributors

Both authors were involved in patient care, reviewing the literature, and writing this case report. Both authors approved the final manuscript.

Conflict of Interest

The authors declare that they have no conflict of interest regarding the publication of this case report.

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Patient Consent

Obtained.

Provenance and Peer Review

This case report was peer reviewed.

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