Rectosigmoid Perforation and Sepsis from Endometriosis in Early Pregnancy

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Abstract

Background: We report diagnosis and management of complicated intra-abdominal infection secondary to rectosigmoid perforation from endometriosis in early pregnancy.

Case Presentation: A 30-year-old primigravid Caucasian female with abdominal pain at 15 wks gestation was found to have a massive intra-abdominal infection on laparoscopy. Initial conservative management in light of a highly desired pregnancy failed, and she required bowel diversion because of rectosigmoid perforation at a site of deep endometriosis. Post-operative course was complicated by loss of the pregnancy and multiple percutaneous drainage procedures.

Conclusion: Bowel perforation as cause of acute abdomen in the setting of endometriosis should be considered in early pregnancy. With a pre-viable fetus, all measures must be taken to save the mother and definitive treatment may require bowel diversion or resection.

ENDOMETRIOSIS is the benign, estrogen-dependent inflammatory proliferation of endometrial glands and stroma at extra-uterine sites and can involve various peritoneal and extra-peritoneal structures [1]. The exact prevalence is not known and many females remain either asymptomatic or undiagnosed. Definitive diagnosis involves direct visualization of endometriotic implants via surgery [1]. Reports of visually confirmed endometriosis range from 1% of females undergoing a major operation for any gynecologic indication to as high as 50% of females undergoing laparoscopy for chronic pelvic pain or infertility [2,3].

Endometriosis may present as deeply infiltrative implants, also known as deep endometriosis. Deep endometriosis is defined as endometriosis infiltrating greater than 5 mm below the peritoneal surface and can be found primarily on the uterosacral ligaments, the rectovaginal septum, vagina, and the rectosigmoid area [4]. Bowel involvement is estimated to occur between 3%–37% of all cases, and in 90% of these cases when the rectum or sigmoid are involved [5]. Endometriosis is classified into one of four stages (I-minimal, II-mild, III-moderate, and IV-severe) depending on the location, extent, and depth of implants; the presence and severity of adhesions; and presence and size of ovarian endometriomas [6].

Symptoms of endometriosis are believed to regress during pregnancy as the hormonal environment produced by pregnancy may inhibit symptoms [6]. However, existing endometriotic implants can persist and progressive traction of the enlarged uterus on adherent implants, coupled with increased intra-abdominal pressure, can result in perforation when these implants reside on hollow viscous organs [7]. Prior reports of perforation have been reported to occur between 20–40 wks of pregnancy [7]. We report the case of a 30-year-old pregnant woman who presented with an acute abdomen and was diagnosed subsequently with a rectosigmoid bowel perforation secondary to deep endometriosis at 15 wks gestation.

Case Presentation

A 30-year-old primigravid Caucasian female with history of stage IV endometriosis presented at 15 wks gestation with worsening abdominopelvic pain. Prior to her pregnancy, she had undergone several laparoscopic procedures with removal of endometriotic implants and lysis of adhesions with the most recent procedure occurring 3 mo prior to achieving pregnancy. In the week prior to admission, she had presented to the emergency department (ED) where a pelvic ultrasound

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FIG. 1. Pre-operative transvaginal ultrasound demonstrating mildly heterogeneous lesion of the left adnexa measuring $7.5 \times 5.3 \times 6.2$ cm with appearance suggestive of endometrioma.

revealed a 6 cm left ovarian cyst as well as an elevated white blood cell count (WBC) of approximately 20 k/uL. She followed up with her obstetrician-gynecologist and a repeat pelvic ultrasound revealed the left ovarian cyst increased to 8 cm in size. On this presentation to the ED, she had significant worsening of her pain and a pelvic ultrasound confirmed the left ovarian cyst (Fig. 1). At this time, a complete blood count revealed WBC elevated to 27.76 k/uL. Physical examination revealed peritoneal signs and she was taken urgently for diagnostic laparoscopy and started on antibiotics. Pre-operatively, the patient stressed that she wished for everything possible to save this very desired pregnancy.

Intra-operative findings included an enlarged uterus consistent with a 15-wk pregnancy, 300–400 ccs of pus in the abdomen (Fig. 2) along with extensive inflammatory adhesions (Fig. 3). The left tubo-ovarian complex was not fully visualized because of dense adhesions but appeared consistent with a hydrosalpinx and the left adnexa could not be visualized. Attempts to separate the sigmoid colon and rectum from the uterus at the pelvic brim resulted in uterine bleeding and were abandoned. The appendix was identified in the inflammatory debris and appeared secondarily inflamed but with no sign of rupture. Intra-operative consultation with general surgery was sought and an appendectomy was performed. Two large bore Jackson-Pratt (JP) drains were placed along the right gutter and over the uterus in the left

FIG. 2. Intra-operative image during diagnostic laparoscopy demonstrating pus in the abdominal cavity and enlarged uterus consistent with 15-wk pregnancy.

FIG. 3. Intra-operative image during diagnostic laparoscopy demonstrating inflammatory adhesions of the omentum to the abdominal wall.

gutter, respectively. In the immediate post-operative period, fetal heart tones were confirmed using Doppler.

The patient was continued on intravenous (IV) antibiotics. She initially improved but quickly exhibited peritoneal signs and became increasingly tachypneic and tachycardic over the next 12 h with rising WBC to 35.58 k/uL. She was transferred to the surgical intensive care unit (SICU) and further resuscitated. Given her clinical de-compensation and findings of peritonitis, she was returned to the operating room (OR) for an exploratory laparotomy with the assistance of colorectal surgery. On re-exploration, a large abscess, likely representing the “ovarian cyst” observed on prior ultrasound, was drained in the left pelvic sidewall. A site of presumed visceral perforation was identified in the distal sigmoid colon. Intra-operative flexible sigmoidoscopy revealed purulent drainage within the rectum and a sinus tract within an endometrioma at the site of presumed perforation. An air leak test was performed by insufflating the colon under saline. The perforation had sealed seemingly as there was no leakage of air through the perforation site. Dense adhesions prevented safe dissection of the sigmoid off the uterus. A diverting loop ileostomy was performed to provide source control of the perforation and prevent ongoing sepsis while preserving the gravid uterus and the two JP drains continued. Post-operatively, the patient reported significant improvement of her pain. Approximately 24 h after the second operation, she experienced spontaneous rupture of membranes with subsequent fetal death and underwent misoprostol induction of labor. She was discharged home the following day in stable condition to continue 2 wks of antibiotics. She required subsequently placement of additional percutaneous drains because of recurrence of the intra-abdominal abscess. Ultimately, her final drain was removed 8 wks following her definitive operation and she recovered fully. At this time, she continues to explore options to preserve her fertility before definitive surgical treatment of her endometriosis, as definitive surgery may require both hysterectomy and en bloc bowel resection.

Discussion and Literature Review

In this case, we described the management of a gravid female who was found to have massive intra-peritoneal infection secondary to bowel perforation at the site of deeply infiltrative endometriosis. This case is unique in that the complication occurred at 15 wks gestation with a pre-viable fetus. On initial presentation, the patient made it known that this pregnancy was highly desired in the setting of stage IV

endometriosis. After admission, laparoscopy clearly confirmed intra-peritoneal infection, but the source remained unclear, and initial surgical efforts were limited out of concern for disrupting the pregnancy but was ultimately unsuccessful. In the setting of an acutely decompensating patient and a pre-viable fetus, all subsequent efforts were appropriately turned to saving the patient. It was only with identification of bowel perforation and definitive treatment with bowel diversion that the patient began to improve.

Although endometriosis improves typically during pregnancy, the current case demonstrates the potential occurrence of serious and unexpected complications of the disease. Additionally, patients with deep endometriosis may not have a history indicating increased risk of bowel perforation during pregnancy. This described patient did not have a history of bleeding per rectum during menses and had undergone a pre-conception colonoscopy with no specific findings. Additionally, although a pre-conception diagnostic laparoscopy did reveal the rectosigmoid colon to be densely adherent to the posterior uterus, this was believed to be a frequent finding in the setting of severe disease and not a clear indication for preconception bowel surgery [4]. Bowel perforation linked to endometriosis during pregnancy has been described in the literature with a majority of cases occurring in the third trimester of pregnancy [7]. To our knowledge, however, perforation as early as 15 wks gestation has not been reported and highlights the fact that this type of complication can occur relatively early in pregnancy and should remain on the differential when contemplating causes of acute abdomen in the pregnant patient. Precise diagnosis must be made about the location and extent of the injury. Although ultrasound is the predominant imaging modality used in pregnancy, it may not be the most accurate modality to diagnose bowel perforation. Computed tomography (CT) has been shown to be accurate for predicting the site of gastrointestinal perforation [8]. Diagnostic CT imaging typically exposes a fetus to less than 0.05 Gy of ionizing radiation and although there has been no convincing evidence of increased risk of fetal anomalies, intellectual disability, growth restriction, or pregnancy loss with use of CT, widespread hesitancy still exists for utilizing CT during pregnancy [9]. Given the risk of fetal and maternal death with undiagnosed intra-abdominal sepsis, CT should be employed for accurate diagnosis in cases of diagnostic ambiguity. Once an accurate diagnosis is made on the location, size of the lesion, and site of involvement, full thickness excision, bowel diversion, or bowel resection may be performed as the standard of care [8]. In the OR, intra-operative colonoscopy or flexible sigmoidoscopy can be utilized as an accurate method to further localize a bowel perforation [10].

Conclusion

In summary, despite its relative rarity, hollow viscous perforation secondary to deep endometriosis of the bowel must be considered as a cause of acute abdomen in pregnancy, and will ultimately require surgical intervention with either resection or diversion. It must also be noted that this serious complication can occur essentially any time in the pregnancy, and that appropriate management may cause ultimately loss of the pregnancy. The result of proper management is a prompt reversal of most morbidities for affected females, though depending on the stage and extensiveness of

endometriosis, further multi-disciplinary treatment may be necessary prior to complete return to normalcy.

Author Disclosure Statement

The authors have nothing to disclose.

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Abbreviations Used

CT = computed tomography

ED = emergency department

IV = intravenous

JP = Jackson-Pratt

OR = operating room

SICU = surgical intensive care unit

WBC = white blood count

Cite this article as: Chang ES, Bateman BG, Hedrick TL, and Modesitt SC (2016) Rectosigmoid perforation and sepsis from endometriosis in early pregnancy, *Surgical Infections Case Reports* 1:1, 32–34, DOI: 10.1089/crsi.2016.0005.