Bowel complications of deep endometriosis during pregnancy or in vitro fertilization

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Objective: To review bowel complications caused by deep endometriosis during pregnancy or in vitro fertilization (IVF).

Design: Three case reports and a systematic review.

Setting: A tertiary referral center for deep endometriosis surgery.

Patient(s): Three case reports of bowel perforation or occlusion during pregnancy caused by deep endometriosis.

Intervention(s): A PubMed search was conducted to identify complications of deep endometriosis during pregnancy or IVF. The literature search identified 13 articles. According to these, 12 articles described 12 bowel complications caused by progression of deep endometriosis during pregnancy, and 1 article described six cases of bowel occlusion during IVF.

Result(s): In 12 of 15 women, complications occurred during the third trimester of pregnancy, whereas 3 of 15 women presented with complications in the postpartum period. All complications during IVF occurred during stimulation. No specific factors that could predict these complications were identified, leading to the conclusion that endometriosis complications that occur in pregnancy or in IVF patients are probably underreported.

Conclusion(s): Bowel complications during pregnancy or IVF stimulation may occur in women with deep endometriosis. This suggests

that the endocrine environment of pregnancy does not prevent progression, at least in some women. These complications are rare, although probably underreported. (Fertil Steril® 2014;101:442–6. ©2014 by American Society for Reproductive Medicine.)

Key Words: Endometriosis, pregnancy, IVF, bowel perforation, bowel occlusion

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ndometriosis is defined as the presence of glands and stroma outside the uterus. It is an enigmatic disease, and its pathophysiology, progression, and natural history are poorly understood. Even though it is believed to be a progressive disease, progression was not seen in some women followed for more than 1 year without surgery (1). The hormonal responsiveness of the lesions is believed to be comparable to that of the endome-

trium. Nevertheless, it is unclear whether the hormonal responsiveness of all deep endometriosis lesions follows a similar pattern. Indeed, it is still unclear whether the endometriotic cells are identical to the endometrium, as suggested by the Sampson theory of retrograde menstruation and implantation. If, however, deep endometriotic lesions are the consequence of a genomic incident (2), the hormonal responsiveness of each lesion might be different.

During pregnancy, superficial endometriosis lesions have been reported to decidualize, preventing progression. The use of pseudopregnancy or oral contraceptives as a medical treatment for endometriosis (3) is, indeed, based on this mechanism. For deep endometriosis, however, decidualization during pregnancy has, to the best of our knowledge, not yet been confirmed.

Three patients presenting recently to our center with deep endometriosis causing a spontaneous bowel perforation during pregnancy prompted us to review the literature.

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MATERIALS AND METHODS

A systematic review was performed using the PRISMA (Paris Rome Italy States Mother Africa) guidelines. A

combination of the keywords "pregnancy," "endometriosis," "IVF," "bowel perforation," "intestinal perforation," and "intestinal occlusion" was used to identify the maximum number of relevant citations in PubMed. Institutional review board approval was not required for such a literature review or the case reports.

RESULTS

Systematic Review

In the literature, 12 case reports of bowel perforation during pregnancy caused by endometriosis were found: 2 involved the small intestine, 1 the caecum, 3 the appendix, and 6 the rectosigmoid colon (4–15). Seven of twelve perforations occurred between the 26th to the 37th weeks of pregnancy (information not available in 2 cases) and the remaining three in the immediate postpartum period. All perforations presented as an acute abdomen, and the pathology reported deposits of endometriosis on the bowel. All patients underwent emergency surgery for acute diffuse peritonitis, and a Hartman operation and/or segmental resection was performed. Healthy babies were delivered in all seven cases; a cesarean section was performed in four of seven cases, whereas in three of seven cases the baby was delivered by vaginal birth. Surprisingly, only 3 of 12 women had a clear history of endometriosis, whereas in 4 of 12 such information was missing and the remaining 5 of 12 had no history.

Six case reports of bowel occlusion during IVF were reviewed. In all cases a rapid progression of a small sigmoid lesion was observed (16). All patients had been diagnosed with a stage IV endometriosis. Three patients had undergone excision of endometriotic lesions before ovarian stimulation.

Case Reports

Written informed consent was obtained from the patients for the publication of these case reports.

Case 1. A 36-year-old Portuguese woman with one uneventful pregnancy was diagnosed with a 3-cm rectosigmoid endometriosis nodule by magnetic resonance imaging. Both the mother and sister also had deep endometriosis. Because of infertility, the patient underwent IVF treatment and became pregnant during the ninth cycle. During the 28th week of pregnancy she presented with acute and severe abdominal pain. Pyelone-phritis was diagnosed, and *broad-spectrum antibiotics* were initiated, but her symptoms got worse and she became septic. An exploratory laparotomy confirmed the peritonitis with large quantities of purulent liquid. A bowel perforation, however, was not found, although cultures revealed *Escherichia coli*. With antibiotic treatment the patient gradually improved and was discharged 2 weeks later. At 37 weeks of pregnancy, a healthy baby was delivered by cesarean section.

The patient did not breast feed, and 2 months later she returned with complaints of dysmenorrhea, severe pelvic pain, catamenial diarrhea, rectorrhagia, and dyschesia.

At clinical examination a 4-cm rectovaginal nodule invading the vagina and a 2-cm bladder nodule were identified.

Colonoscopy revealed a third endometriotic nodule in the sigmoid causing severe stenosis (<2 cm lumen). Magnetic

resonance imaging diagnosed cystic ovarian endometriosis (a 3-cm and a 4-cm nodule on the right and left ovary, respectively) and bilateral hydrosalpinx.

The patient underwent total laparoscopic hysterectomy, adnexectomy, partial cystectomy, rectovaginal septum nodule excision, and segmental resection of the rectosigmoid colon by laparoscopy. Pathology confirmed the diagnosis of endometriosis.

Throughout the follow-up the patient was completely asymptomatic.

Case 2. A 35-year-old Portuguese woman with one uneventful pregnancy and without any relevant medical or family history presented with severe pain symptoms suggestive of deep endometriosis. She became spontaneously pregnant, and at the 35th week of pregnancy she experienced acute and severe abdominal pain for which a laparotomy was performed. A healthy baby was delivered by cesarean section, during which a concealed bowel perforation was detected and thus a rectosigmoid segmental resection with latero/lateral anastomoses as well as appendectomy was performed.

The pathology report revealed colonic, nodal, and appendicular endometriosis with pseudodecidualization of the stroma.

Six months later the patient presented with dyspareunia, and a 3-cm rectovaginal nodule with vaginal invasion was clinically detected. A vaginal ultrasound scan confirmed the presence of deep endometriosis involving the rectovaginal septum and the sigmoid infiltrating the muscularis of the rectum.

Because she was breast feeding without severe pain symptoms, expectant management with oral contraceptives was chosen. Colonoscopy performed 6 months after delivery revealed no residual disease. The patient's quality of life remained satisfactory throughout the 6-month follow-up.

Case 3. An asymptomatic 34-year-old Portuguese woman without symptoms suggesting endometriosis was admitted with severe abdominal pain and pollakisuria during the 16th week of a spontaneous pregnancy. A vaginal ultrasound scan detected a septated hemorrhagic cyst of 11×10 cm of the left ovary and free peritoneal fluid. A laparotomy was performed because torsion was suspected. During surgery, multiple pelvic abscesses were found involving the uterus, the left ovary, the sigmoid colon, and the small bowel. The capsule of the ovarian cyst was broken. After lavage the patient remained in intensive care and underwent multiple peritoneal rinsings until resolution of peritonitis. Abdominal wall closure was performed 6 days later.

Ten days later the patient deteriorated, with symptoms of bowel occlusion and sepsis. A second laparotomy was performed, and several abscesses were detected. Forty-eight hours later a Hartman's procedure was performed for diffuse peritonitis caused by sigmoid perforation. Two more lavage revisions by laparotomy were necessary, and 20 days later the wall was closed. Bacteriologic tests reported *E. coli*, whereas the pathology report showed a "mucosal perforation due to decidualization of endometriotic lesion" in the resected bowel specimen.

The patient underwent an uneventful vaginal delivery at 39 weeks, but because she never returned for consultation, no follow-up is available.

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