

Rectal perforation from endometriosis in pregnancy: Case report and literature review

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consistent with decidualization of the rectal wall. Only 20 cases of intestinal perforation due to endometriosis have been reported in the literature. This report is believed to be the first case of spontaneous rectal perforation from endometriosis in pregnancy, and it shows the potential occurrence of serious and unexpected complications of the disease.

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Abstract

This case report describes a woman with spontaneous rectal perforation from decidualized endometriosis in pregnancy. A 37-year-old woman was admitted to our hospital at 30 wk of pregnancy with symptoms suggestive of pyelonephritis, which persisted until 33 wk of gestation when delivery of a premature male baby was performed through a cesarean section. On postoperative day 2, an abdominal computed tomography showed free air in the peritoneal cavity and a pelvic abscess. Explorative celiotomy revealed a diffuse severe fecaloid peritonitis that originated from a 3-cm wide rectal perforation. A Hartmann operation was then performed. Histopathological findings were

INTRODUCTION

Endometriosis is defined by the presence of endometrium outside the uterus and usually affects pelvic structures including the bowel. Intestinal involvement occurs in 3%-37% of patients with endometriosis^[1]. Intestinal endometriosis typically takes the form of asymptomatic serosal implants that occasionally result in intestinal obstruction with recurrent abdominal pain^[2]. Transmural involvement is not as common, and spontaneous perforation of intestinal endometriosis is a rare complication that generally occurs in pregnancy^[2-4].

We report the case of a pregnant woman with spontaneous rectal perforation that originated from a decidualized endometriotic nodule, and we review all cases of intestinal perforation from endometriosis reported in the literature. To the best of our knowledge, this is the first report of spontaneous perforation of the rectal wall in an endometriotic area in pregnancy, which is an extremely rare cause of acute abdomen.

CASE REPORT

In October 2008, a 37-year-old Caucasian woman was admitted to our hospital because of colicky pain in the right iliac fossa at 30 wk of pregnancy. She had a previous history of endometriosis that was surgically treated. In 2003, she underwent a laparotomy excision of an endometrioma of the left ovary. Later, she was submitted to three subsequent laparoscopies in which left salpingectomy for a sactosalpinx, diathermocoagulation of multiple endometriotic foci, excision of the posterior vaginal fornix, and adhesiolysis mainly between the uterus and sigmoid colon were performed. In 2007, the final laparoscopy showed no endometriosis in the Douglas cul-de-sac. At admission to our hospital, abdominal ultrasound revealed pelvicalyceal dilation on the right side, which improved after double J stent placement. However, symptoms suggestive of pyelonephritis persisted until 33 wk of gestation when delivery of a 1600 g premature male baby was performed through a cesarean section, because of the mother's worsening clinical condition that was caused by developing sepsis that was apparently related to pyelonephritis. Indeed, the clinical diagnosis of pyelonephritis was also supported by detection of pathological levels of *Escherichia coli* in the urine. However, during cesarean section, at the moment of peritoneal incision, a fecaloid smell came from the peritoneal cavity, but unfortunately, this finding was not correctly understood. On postoperative day 2, the patient was pyrexial (40°C), pale and shocked. Blood pressure and pulse rate were 90/50 mmHg and 95 beats/min, respectively. The abdomen was distended, with tenderness and rebound in the lower quadrants, where bowel sounds were absent. Laboratory data were as follows: white blood cell count, $204 \times 10^9/L$; red blood cell count, $370 \times 10^{12}/L$; hemoglobin, 10 g/L; thrombocyte count $461 \times 10^9/L$; fibrinogen, 6.54 g/L. Chest X-ray revealed bilateral pleural effusion, while abdominal plain radiography showed free air in the peritoneal cavity. Enhanced abdominal computed tomography (CT) confirmed these findings (Figure 1A), as well as a low-density area with the features of a pelvic abscess on the right side of the enlarged uterus (Figure 1B). The patient underwent an emergency operation for a presumptive diagnosis of acute diffuse peritonitis. Explorative celiotomy revealed diffuse, severe fecaloid peritonitis that originated from a 3-cm wide rectal perforation in the deep cul-de-sac. Diffuse peritonitis was in an advanced stage and lasted for > 48 h. In addition, the sigmoid colon was adher-

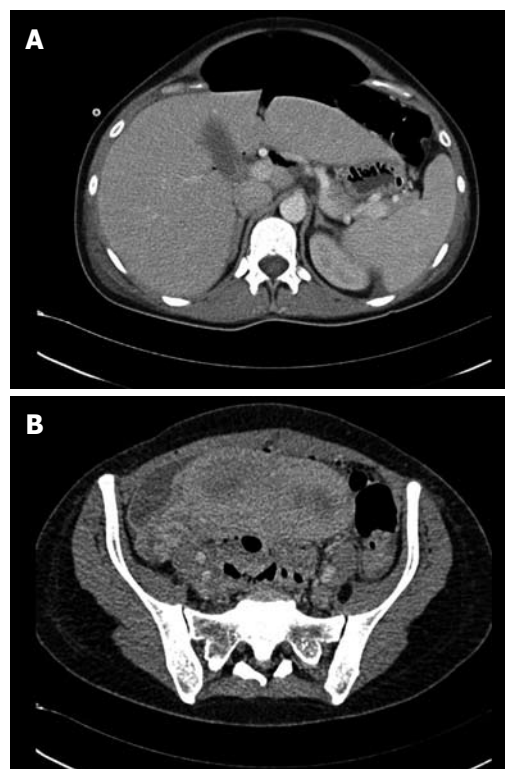


Figure 1 Axial computed tomography (CT) scan. A: Free air in the abdominal cavity; B: Pelvic abscess at the right side of the enlarged uterus.

ent to the uterus. A Hartmann operation was necessary with end colostomy using the descending colon and closure of the rectal stump, and appendectomy was performed. Bilateral pleural effusion persisted for 14 d after the operation and the patient was finally discharged on postoperative day 19 as a consequence of abdominal wound infection. On gross examination, rectal perforation seemed to originate from an endometriotic nodule. Histopathological findings were consistent with decidualization of the rectal wall (Figure 2A) and vermiform appendix. Decidualized endometriosis was mainly found around the rectal perforation, which was the most likely explanation for this occurrence (Figure 2B). Nursery care was needed for 35 d until the infant reached a stable body weight and was able to feed by mouth. Afterwards, the patient was referred to the gynecologist for further therapy and she was readmitted 3 mo later for Hartmann reversal without any sequelae.

DISCUSSION

Intestinal endometriosis may affect the ileum, appendix, sigmoid colon and rectum^[4]. The most common non-genital manifestation is in the rectosigmoid^[3,5]. The peritoneal implantation of endometrium by retrograde menstruation or the possible metaplasia of peritoneal cells are still the two most accepted etiological theories of endometriosis^[1].

Intestinal endometriosis may be found in every layer of the bowel wall but it is most commonly found within the

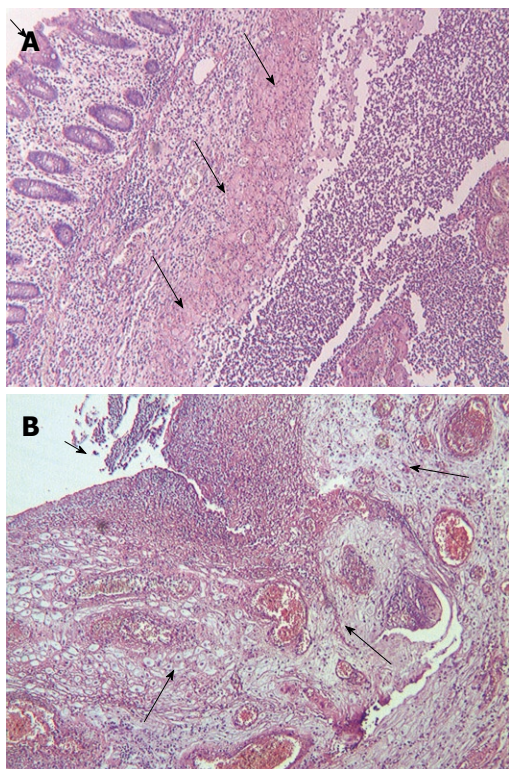


Figure 2 The rectal wall. A: Decidualization of the rectal wall (long arrows); mucosa side of the rectal wall (short arrow) (HE, × 40); B: Decidualized endometriosis around the rectal perforation (long arrows); rectal perforation with necrosis at the peritoneal side of the rectal wall (short arrow) (HE, × 100).

subserosa as superficial serosal implants^[2,6]. Under cyclical hormonal influences, these implants may proliferate and infiltrate the intestinal wall and cause a fibrotic reaction with formation of strictures and adhesions, which may lead to bowel obstruction and recurrent abdominal pain^[4,7]. On the other hand, transmural bowel wall involvement is not so common and the intestinal mucosa usually remains intact, and perforation of the affected intestinal tract is a very rare complication^[3,6]. Up till now and including the present report, only 21 cases of intestinal perforation from endometriosis have been reported and among these, nine cases (42.8%) occurred in pregnancy. Sigmoid colon turned out to be the most common site of perforation from endometriosis and accounted for 12 of 21 published cases. The current case is the first report of spontaneous perforation of the rectum (Table 1).

In those patients with perforation, the entire intestinal wall is replaced by endometriotic tissue. In pregnancy, under the effect of progesterone, the area of ectopic endometrium becomes decidualized with a progressive reduction in size^[8,9]. In our patient, decidualized endometriosis was found mainly around the rectal perforation. Actually, the reduction in size of a transmural endometriotic nodule may lead to perforation, by weakening of the intestinal wall^[2], particularly in the third trimester, which is the time of perforation in most reported cases^[2,8], as in our patient who had rectal perforation at 33 wk of pregnancy. Moreover, decidualization causes a severe inflammatory response with an increased number of natural killer cells

Table 1 Literature review of intestinal perforation from endometriosis

Yr	Author	Journal	Site of perforation	n	Pregnancy
1931	Haufler ^[11]	<i>Virchows Arch</i> [in German]	Jejunum	1	Yes
1955	Henriksen ^[12]	<i>Am J Surg</i>	Sigmoid colon	1	No
1977	Clement ^[13]	<i>Br J Obstet Gynaecol</i>	Sigmoid colon	1	Yes
1979	Rud ^[14]	<i>Ugeskr Laeger</i> [in Danish]	Sigmoid colon	1	Yes
1981	Gini <i>et al</i> ^[15]	<i>Br J Obstet Gynaecol</i>	Vermiform appendix	1	Yes
1984	Floberg <i>et al</i> ^[16]	<i>Acta Obstet Gynecol Scand</i>	Sigmoid colon	1	Yes
1987	Nakatani <i>et al</i> ^[17]	<i>Acta Pathol Jpn</i>	Vermiform appendix	1	Yes
1988	Strömberg <i>et al</i> ^[18]	<i>Lakartidningen</i> [in Swedish]	Sigmoid colon	1	No
1988	Ledley <i>et al</i> ^[19]	<i>Am J Gastroenterol</i>	Sigmoid colon	1	No
1990	Goodman <i>et al</i> ^[20]	<i>Gastrointest Radiol</i>	Sigmoid colon	1	No
1992	Bakri <i>et al</i> ^[21]	<i>Int J Gynaecol Obstet</i>	Sigmoid colon	1	No
1993	Yelon <i>et al</i> ^[22]	<i>J Clin Gastroenterol</i>	Vermiform appendix	1	No
1994	Allimant <i>et al</i> ^[23]	<i>J Chir (Paris)</i> [in French]	Ileum	1	No
1995	Abbo <i>et al</i> ^[24]	<i>Minerva Chir</i> [in Italian]	Sigmoid colon	1	No
2000	Bossotti <i>et al</i> ^[25]	<i>Chir Ital</i>	Ileum	1	No
2004	Decker <i>et al</i> ^[3]	<i>Arch Gynecol Obstet</i>	Ileum	1	No
2006	Schweitzer <i>et al</i> ^[4]	<i>Int J Gynaecol Obstet</i>	Sigmoid colon	1	Yes
2008	Faucheron <i>et al</i> ^[9]	<i>Colorectal Dis</i>	Vermiform appendix	1	Yes
2008	Shaw <i>et al</i> ^[26]	<i>Colorectal Dis</i>	Sigmoid colon	1	No
2009	Garg <i>et al</i> ^[2]	<i>World J Gastroenterol</i>	Sigmoid colon	1	No
2010	Pisanu <i>et al</i>	<i>Present report</i>	Rectum	1	Yes
			Total	21	9 Y/12 N

and decidual changes, which are responsible for a higher risk of perforation^[9,10]. In our case, we believe that perforation was also facilitated by the progressive traction of the enlarged uterus on the strictly adherent sigmoid colon, and consequently, on the decidualized and weakened area of the anterior rectal wall. Moreover, the absence of endometriotic foci in the cul-de-sac at final laparoscopy made rectal perforation unpredictable.

Although endometriosis improves during pregnancy, the current report shows the potential occurrence of serious and unexpected complications of the disease. Both the rareness of the perforation and the symptoms that are suggestive of pyelonephritis or diverticulitis may be misleading and delay the diagnosis. Indeed, the appropriate management of these patients may be challenging and a good outcome is absolutely dependent on a multidisciplinary approach.

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