

Postpartum complications in a patient with a previous proctocolectomy and ileo-pouch-anal anastomosis (IPAA) for ulcerative colitis

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Summary

This case report regards pregnancy and delivery of a patient who had undergone proctocolectomy and ileo-pouch-anal-anastomosis (IPAA) for ulcerative colitis. The patient delivered through caesarean section and experienced serious complications postpartum. Such complications have been described in association with Chron's disease and have never been described after proctocolectomy and IPAA for ulcerative colitis. This case report suggests that the limit between these two diseases is not sharp.

Key words: Ileo-pouch-anal-anastomosis, pregnancy, delivery, complications, ulcerative colitis.

Introduction

Inflammatory bowel disease (IBD) is a chronic disease affecting mainly young people in their reproductive years. This article describes the case of unusual puerperal complication following caesarean section in a patient, who had undergone proctocolectomy and ileo-pouch-anal-anastomosis for ulcerative colitis.

Case report

A 28 year old woman underwent proctocolectomy and ileostomy in May 1998, and Ileo-Pouch-Anal-Anastomosis (IPAA) in November 1998, due to ulcerative colitis.

In May 1999 the woman, gravida 1, para 0, came to us for obstetric care at 7 weeks' gestation. Her clinical condition was good, her weight was 52 kg, her blood pressure was 100/60mmHg. She was given iron and folic acid due to mild anaemia; and mineral salts with vitamins for the reduced intestinal absorption. Serial ultrasound scans showed regular fetal growth (although at the middle-lower end of the normal range expected for gestational age) and normal morphology. Amniocentesis was performed: the karyotype was 46, XX. Apart from two episodes of enteritis, the function of IPAA was always good. At 25 weeks' gestation serum titres of anti-Cardiolipine antibodies were slightly increased: acetylsalicylic acid was administered until 33 weeks, when they returned within the normal range. The course of pregnancy was uneventful until 36 weeks' gestation, when a premature labour started. At this time the maternal clinical condition was good, her weight was 66 kg (a 14 kg gain), her blood pressure was 100/60mmHg. On recommendation of the surgeon who had previously operated on the patient, a caesarean section (C.S.) was performed under general anaesthesia and antibiotic prophylaxis with ceftriaxone. A female infant weighing 2550g, with Apgar scores 9/10, was delivered. An interesting surgical finding was an important conglomeration of ileal loops adherent to the right portion of the uterine fundus. Upon recommendation of the surgeon who had performed the IPAA intervention previously, no surgical manipulation was performed on intestine.

On postoperative day 2, after she passed stools, severe abdominal pain and abdominal distension without peritonism appeared. Such symptoms spontaneously disappeared within 24 hours. The rest of her postoperative course was uneventful, apart from puerperal blues, lack of appetite and a mild transient fever attributed to lactogenesis on postoperative day 5, in spite of antibiotics. Mother and baby were well on postoperative day 7, when they were discharged and antibiotics were discontinued. On the 8th day postpartum, fever recurred: therefore, antibiotic therapy with oral ciprofloxacin was started. Furthermore, abdominal pain and distension, without peritonism, occurred under therapy with methyl-ergometrine and ceased following its discontinuation. Lactation was inhibited with cabergoline, due to the poor amount of maternal milk. On the 9th day postpartum, while persisting the fever, a severe pain appeared at the basis of the left hemithorax: a chest-X-ray confirmed the clinical suspect of a left pleural effusion, and revealed free gas under the right hemidiaphragm, which was at first attributed to the C.S. The patient was hospitalised again, oral ciprofloxacin was discontinued and a therapy with intravenous piperacilline and indomethacine resulted in resolution of

fever and pain three days later. On the 13th day postpartum, however, an entero-cutaneous fistula became evident after the patient had a breakfast and without abdominal distension, pain or peritonism. Total parenteral nutrition via a central catheter (approximately 3000 Kcal.) was started. Chest and abdominal radiography revealed the resolution of the pleural effusion, and confirmed the previously detected gas collection suggesting now a sackful perforation, which prompted addition of metronidazole and amikacine in therapy. The fistula was silent, but fever recurred, becoming swinging and reaching over 41°C on the 18th day postpartum: then, a thoraco-abdominal computed tomography (CT) scan revealed a fluid collection above and behind the spleen, confirming free gas above the liver (Fig.1). At laparotomy, a plastic peritonitis made intestinal exploration impossible; a subdiaphragmatic, retro splenic and partially retrohepatic abscess of approximately 800 cc was drained, the residual cavity was washed and two drains were left in situ; a perforation of the "pouch" was excluded endoscopically. Postoperative course was uneventful, parenteral nutrition was discontinued on postoperative day 5, and the Patient discharged on postoperative day 7. The patient is still under follow-up at the Gastroenterology Unit, where the ulcerative colitis had been diagnosed and treated. She has been well until 2009, when an entero-vaginal fistula became evident: fistulography revealed that it was actually a pouch-vaginal fistula. The fistula was not treated surgically; it resolved after ten months therapy with oral ciprofloxacin (cycles of ten days every month at a dose of 1000 mg daily)



Figure 1. CT scan of the upper abdomen showing a left sub-diaphragmatic fluid collection with an air-fluid level, and free gas under the right hemidiaphragm.

Discussion

Indication to C.S. in patients with IPAA does not emerge from the world literature and vaginal delivery may be considered in these cases (1). However, in our case, the indication to C.S. was specifically recommended by the surgeon who had performed the IPAA, since the risk of dehiscence of the anastomosis was evaluated as high, also because the previous intervention was recent. The impossibility to explore the bowel at surgery did not allow to clarify with certainty the pathogenesis of complications observed in this unusual case. However, in retro-

spect, the most likely hypothesis for their pathomechanism is represented by the occurrence of one or more ileal perforations, resulting in an entero-cutaneous fistula. The entero-cutaneous fistula, the pneumoperitoneum evidenced radiologically, the plastic peritonitis, and the abscess (which might also have been related to the previous interventions) suggest such a pathogenesis. Maybe, the retraction of the uterus following the C.S., avulsing the wall of the ileal loops adhered to its fundus (as observed at surgery), might have played a role in causing perforations: this is also suggested by the onset of severe abdominal pain and distension under therapy with methylergometrine in the postpartum period.

Whatever the pathogenesis, to our knowledge, the complications reported here have never been described in association with pregnancy and delivery of a woman who had undergone proctocolectomy and IPAA for ulcerative colitis, since this pathology does not recur after proctocolectomy.

Conversely, these are classical complications, in various associations with each other, of Crohn's disease. In fact, various types of enteric fistula, bowel perforations, abdominal abscesses (especially after previous interventions) and even septic shock have been reported during pregnancy or after delivery in Patients with Crohn's disease (2-6). In particular, Solomon et al. (1996) described the avulsion of the ileal wall by the rapid retraction of the uterus in the postpartum period as the probable pathogenesis of an ileal perforation in their case report of exacerbation of Crohn's disease in pregnancy (3). This should be taken into account, when considering the opportunity and dosages of postpartum uterotonic therapy in these Patients.

Moreover, preterm labour, observed in our case, is described as a possible complication in pregnancies of patients with either Crohn's disease or ulcerative colitis (7,8). However, the incidence of such a complication in Crohn's disease is reported to be significantly higher than in ulcerative colitis (8).

Finally, a pouch-vaginal fistula is reported as uncommon complication after IPAA for ulcerative colitis (9,10). Alternative pathogenesis for this complication is also suggested by the occurrence of fistula ten years later the IPAA intervention.

Conclusion

Our case report suggests once again that the limits between ulcerative colitis and Crohn's disease are not sharp and that the Anglo-Saxon term "inflammatory bowel disease" to indicate both pathologies is fully justified.

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