

Case Report

Laparoscopic management for ruptured endometrial cyst in a mid-term pregnancy woman - a case report and literature review

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Abstract: We present a special case of spontaneous endometrial cyst rupture in a mid-term pregnancy and a review of the literature with regards to etiology, diagnosis, and treatment. The patient, at 18 + 5 weeks gestation, was admitted to the Gynecology ward following acute lower right abdominal pain for one week. A right-sided ovarian cyst was diagnosed one year previously. Laparoscopic diagnosis and management of the endometriosis cyst rupture was carried out successfully with no surgical or obstetric complications. The postoperative pathology diagnosis revealed a benign endometrial cyst. Endometrial cyst rupture is a rare complication in pregnancy, which can severely threaten the safety of mother and fetus. Laparoscopic surgery has been extensively applied in the diagnosis and treatment of many causes of acute abdomen pain during pregnancy in recent years.

Keywords: Endometrial cyst rupture, pregnancy, laparoscopic surgery

Introduction

Pelvic endometriosis is a common disease in women of reproductive age, and endometrial cyst rupture is a clinical emergency. However, endometrial cyst rupture is a rare complication during pregnancy. The blood or chocolate-like liquid inside the cyst will flow into the pelvic cavity, which leads to many complications that severely influence the pregnancy outcomes, such as hemoperitoneum, chemical peritonitis, abortion, preterm labor, infection, etc. For patients with endometrial cysts, cystectomy is an advisable choice before pregnancy, to prevent cyst rupture during pregnancy. If acute abdominal pain occurred in pregnancy women due to cyst rupture, laparoscopic exploration is a reasonable approach according to gestational age, status of patient, and the individual doctor's technique. The principle of surgery in pregnancy is to make all efforts to reduce the migration of the uterus and to decrease the risk of abortion or premature birth. Laparoscopic exploration mainly aims to determine the diagnosis or to exclude malignancy. It does not

demand complete lesion resection, but lesion biopsy or cauterization can be applied. Therefore, laparoscopic surgery is of great importance for improving maternal and fetal outcomes in pregnancy women with endometrial cyst rupture.

Case summary

A 28-year-old woman, gravida 2, para 1, at 18 + 5 weeks gestation, was admitted to the gynecology ward at Peking University First Hospital on May 17th, 2017 with a one-week history of lower right abdominal pain. This was on a background of a 5 cm right-sided ovarian cyst, which was identified on physical examination one year previously. At the time, the patient had no abdominal pain, abdominal distention or dysmenorrhea, and no surgical treatment was carried out.

LMP was Jan. 5th, 2017 and was regular prior to this. Self-testing of urine HCG was positive at six weeks. Ultrasonography at seven weeks suggested early intrauterine pregnancy with a

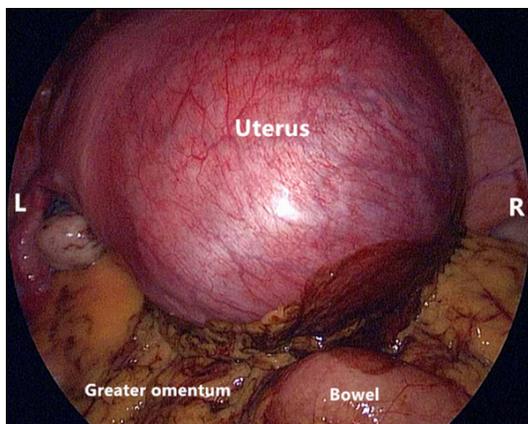


Figure 1. Laparoscopic view of abdominal. The uterus was extremely enlarged, consistent with 19 weeks of gestation, and with no contraction or injury. Crineous chocolate-like liquid had scattered on the surface of greater omentum, peritoneum surface, and pelvic cavity.

live embryo and a 52 × 47 mm right-sided ovarian cyst. The patient had regular antenatal care at the local hospital until one week ago, when she experienced sudden onset lower right abdominal pain. There was no associated vomiting, fever, vaginal bleeding, or contraction of the uterus. After presenting to the local hospital, physical examination and ultrasound scan showed a live fetus at mid-pregnancy, a small amount of ascites, which was interpreted as right-sided ovarian cyst rupture and bleeding. CA-125 was 1010.00 U/ml. The patient was given tocolysis, progesterone, and anti-inflammatory treatment, which led to relief from abdominal pain. Subsequently, the patient consulted our hospital for further diagnosis and treatment. Gynecological ultrasonography revealed an intrauterine live fetus, and a well-defined 29 × 22 × 13 mm hypoechoic cystic area was detected in the right ovary. One week after the onset of pain the CA-125 was 450 U/ml. The possibility of right-sided ovarian endometrial cyst rupture was highly suspected, and as the patient was in stable condition at present with no sign of threatened abortion, conservative treatment was initially recommended. However, the patient had a high CA-125 level and expressed concern regarding malignant transformation. Therefore, as per patient request, a mid-pregnancy laparoscopic exploration was performed under general anesthesia. Crineous chocolate-like liquid, scattered on the surface of greater omentum and bowel in the

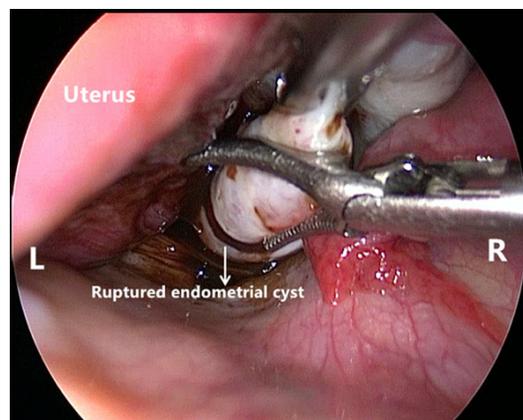


Figure 2. Ruptured endometrial cyst. The right-side ovarian surface was crineous in color and contained a 3 × 2 cm cyst. It had formed dense adhesions with the uterus, bowel, and peritoneum.

pelvic cavity, was seen intraoperatively. The uterus was enlarged consistent with 19 weeks gestation, had regular morphology, and showed gestational period change. There was no contraction or bleeding (**Figure 1**). The right-sided accessory was located in the lower right part of uterus posteriorly. The right-sided ovarian surface was crineous in color, and a 3 × 2 cm cyst was visible inside the ovary. It had formed dense adhesions with the right-sided uterine wall, rectum, peritoneum, and tube, with no active bleeding, papillary nodule, or other visible features suggestive of malignancy (**Figure 2**). Adhesion-lysis and cystectomy was performed, with a wash-out of the chocolate fluid in the cavity. The amount of bleeding was relatively minor, and the operation time is only thirty minutes. Pathology revealed a benign ovarian endometrial cyst. The patient had an unremarkable postoperative recovery, and miscarriage prevention treatment was not given. The patient was discharged as scheduled. Two months later, the ultrasound revealed her fetus, with no structural malformation, was in a good condition and her CA-125 level was 23.5 U/ml.

Discussion

Related factors of ovarian endometrial cyst rupture in pregnancy

Ovarian endometrial cyst is also referred to as ovarian chocolate cyst, which results from recurrent hemorrhage of endometrial lesions outside the ovary. It forms adhesions with the

peripheral tissue or invades the deep cortex to form multilocular cysts, with uneven thickness of the cyst wall. 'Chocolate' refers to the accumulation of brown sticky liquid in the cyst. The ectopic endometrium undergoes recurrent hemorrhage during the menstrual period, which accumulates in the cyst, gradually increasing pressure, thus potentially causing spontaneous rupture of the cyst wall. Therefore, such disease is mostly seen during the menstrual period as well as before and after menstruation. The incidence of ovarian endometriosis cyst in pregnancy is about 0.52% [1], the complications of which include pedicle torsion, cyst rupture, and malignant transformation. The incidence of ovarian cyst pedicle torsion or rupture in pregnancy is higher than that in non-pregnancy [2]. Functional cyst rupture in pregnancy is common, while endometrial cyst rupture in pregnancy is quite rare [3]. The exact reason of cyst rupture remains unclear. It is traditionally believed that ovarian endometrial cysts tend to shrink and the tension tends to become lower under the persistent influence of estrogen and progesterone in pregnancy. Moreover, the cyst is associated with adhesion and limitation of motion, thus it is less likely to develop rupture or torsion. Nonetheless, high concentrations of ovarian progesterone in pregnancy promote decidualization of the ectopic lesion, and increase in local pressure causes hemorrhage in the ectopic lesion. The ovarian blood supply will also increase in pregnancy, thus resulting in increased ovarian volume and the formation of cysts. In particular, the increase of uterine volume with gestational age will rapidly elevate the local ovarian pressure, and fetal movement may also increase the risk of cyst rupture.

Diagnosis of ovarian endometrial cyst rupture in pregnancy

Adnexal cyst torsion or rupture in pregnancy is a common cause of acute abdomen in pregnancy. As is reported in the literature, the diagnosis of adnexal cyst in pregnancy can mainly be confirmed through ultrasound, MRI and tumor markers [4]. Most adnexal cysts in pregnancy are detected and diagnosed through conventional ultrasonography in early pregnancy. After endometrial cyst rupture in pregnancy, the blood or chocolate-like liquid in the cyst will flow into the abdominal cavity, which leads to hemoperitoneum and chemical peritonitis. Thus, a series of clinical symptoms will

occur, which frequently manifest as acute lower abdominal pain associated with tenesmus, nausea, and vomiting [5, 6]. Rupture of smaller cysts may display no clinical symptoms or milder symptoms, as the volume of the cyst contents is lower. Acute abdomen will only be induced in the case of rupture of a large cyst, which is similar to non-pregnancy symptoms. Therefore, endometrial cyst in pregnancy is characterized by atypical clinical symptoms and indistinct signs. The early diagnosis of such disease is more difficult than that in non-pregnancy, thus pre-pregnancy and early-pregnancy ultrasonography is of crucial importance to determine the presence of an endometrial cyst. In this case, the patients mainly manifest with acute spontaneous lower right abdominal pain with no rebound tenderness, nausea, or vomiting.

Significance of CA-125 in the differential diagnosis of ovarian endometrial cyst rupture during pregnancy

CA-125 derives from the fetal coelomic epithelium, which is a group of macromolecular glycoprotein that is synthesized and stored in cells. Intercellular junctions and the obstruction of the cell membrane under normal conditions prevents CA-125 from entering the serum, which means it is low or undetectable in the serum of normal subjects. In the case of tissue malignant transformation, CA-125 synthesized in cell will concentrate at the cell margin, which gives rise to local cell membrane depolarization and thus the CA-125 antigen is transported out of the cell. Infiltrative tumor cell can destroy the tissue structure, and CA-125 secreted after the destruction of intercellular junctions and cell membranes will be released into the blood, thus elevating the serum CA-125. The increased CA-125 is related to numerous diseases, such as ovarian cancer, endometriosis and endometrial cancer. And a higher value predicts higher possibility of disease malignant grade. The CA-125 value in endometriosis is positively correlated with disease severity. Endometriosis patients with a CA-125 value of higher than 200 U/ml should be cautious about the possibility of malignancy or cyst rupture. In this case, the CA-125 value was markedly elevated, and the possibility of cyst rupture was highly suspected based on clinical signs and ultrasonographic appearances. The intraoperative findings further confirmed cyst rupture. The

elevated CA-125 levels may be attributed to the absorption of endometrial cells released into the abdominal cavity and into the blood after cyst rupture. At the same time, it is also linked with the abundant production of CA-125 antigen after the ectopic cells stimulate the biochemical anaplasia of mesothelial cells in peritoneal cavity. Furthermore, pregnancy will further elevate CA-125 [7]. Consequently, comprehensive evaluation should be carried out in pregnant women with remarkably higher CA-125 than before based on medical history, clinical manifestation, physical examination, and auxiliary examinations.

Laparoscopic application for ovarian endometrial cyst rupture during pregnancy

Ovarian endometrial cyst seen under laparoscopy mostly locates in the posterior of uterus, which is associated with adhesion, fixation, and inactivity. The principle of surgery in pregnancy is to make all efforts to reduce the migration of the uterus and to decrease the risk of abortion or premature birth. Laparoscopic exploration mainly aims to determine the diagnosis or to exclude malignancy. It does not demand for complete lesion resection, but lesion biopsy or cauterization can be applied. According to most current literature reports, mid-pregnancy, especially between 14-20 weeks gestation, marks the optimal surgical timing of laparoscopy [8-10], which can reduce the risks of abortion and malformation. During this period, the uterus has not greatly enlarged and there is enough space in the abdominal cavity to avoid injury of the uterus and other viscera during laparoscopic surgery. On the other hand, as there is a large volume of amniotic fluid in the mid-term uterus, touching of the uterus intraoperatively has little influence on the fetus, and thus it is associated with a relatively high safety factor. The abortion rate after laparoscopic surgery has been reported in literature to be as high as 12-15% in early-term and 5-5.6% in mid-pregnancy. In this case, endometrial cyst rupture occurred in mid-pregnancy. Although the pelvic adhesions were serious intraoperatively, and we moved the uterus during separation of adhesions, we did not injure the uterus because the volume of the cyst decreased due to rupture. In addition, the amount of bleeding was relatively minor, and the operation time is only thirty minutes. Therefore, no postoperative and obstetric complications developed. Two months later, the

obstetric system ultrasonography revealed no fetal malformation.

Conclusion

Ovarian endometrial cyst rupture is a rare complication with serious consequences in pregnancy women. Laparoscopic surgery in time properly can improve maternal and fetal outcomes significantly.

Disclosure of conflict of interest

None.

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