**Abstract**

**BACKGROUND**

Heterotopic pregnancy coexisting with a giant ovarian cyst is an extremely rare abnormal pregnancy in the case of natural conception. Its incidence has increased significantly as a result of continuous development in assisted reproductive technologies. When this kind of pregnancy occurs, it threatens the continuation of intrauterine pregnancy and the life of the pregnant woman. Early diagnosis and treatment with safe and effective methods are paramount.

**CASE SUMMARY**

A 30-year-old prim gravida at 8w4d gestation age by scan was admitted to our hospital with heterotopic pregnancy and right ovarian cyst. Laparoscopic resection of the ectopic pregnancy was done but the intrauterine pregnancy and ovarian cyst were preserved.

**CONCLUSION**

Approach in a patient with heterotopic pregnancy and giant ovarian cyst is individualized depending on the fertility requirements. We recommend that:

1. If the patient has satisfied parity (no fertility requirement), laparoscopic salpingectomy and the giant ovarian cyst and removal of intrauterine pregnancy.
2. If the patient has fertility requirement (willing to have more children in future), laparoscopic salpingectomy or salpingostomy can be done with the preservation of the intrauterine pregnancy. Serial ovarian cyst aspiration can be done under ultrasound; resection can be done after delivery.

3. Early diagnosis of heterotopic pregnancy by active surveillance during antenatal visits with the use of ultrasound is important avoid catastrophic complications.

**Key Words:** Heterotopic pregnancy; ectopic pregnancy; giant ovarian cyst; case report

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**Core Tip:** Heterotopic pregnancy coexisting with a giant ovarian cyst is an extremely rare abnormal pregnancy in the case of natural conception. It threatens the continuation of intrauterine pregnancy and the life of the pregnant woman. Early diagnosis and treatment with safe and effective methods are paramount. We report a rare case of heterotopic pregnancy complicated by a giant ovarian cyst. Approach in a patient with heterotopic pregnancy and giant ovarian cyst is individualized depending on the fertility requirements. We recommend that:
1. If the patient has satisfied parity (no fertility requirement), laparoscopic salpingectomy and the giant ovarian cyst and removal of intrauterine pregnancy.
2. If the patient has fertility requirement (willing to have more children in future), laparoscopic salpingectomy or salpingostomy can be done with the preservation of the intrauterine pregnancy. Serial ovarian cyst aspiration can be done under ultrasound; resection can be done after delivery.

**INTRODUCTION**

Coexistence of intrauterine and ectopic pregnancy is called heterotopic pregnancy with incidence of 1/100,000 [1]. Rapid development in artificial reproduction technologies (ART) has given rise to the incidence of heterotopic pregnancies and theca lutein ovarian
cyst [2]. Theca lutein ovarian cyst, also known as hyper-reactive xanthinization were previously thought to occur in multiple pregnancies, hypertensive disorders of pregnancy, blood group incompatibility, trophoblastic disorders, oral administration of large amounts of estrogen and in induced ovulation with gonadotropins. In this paper, we retrospectively analyzed a recent case of heterotopic pregnancy combined with a large theca lutein ovarian cyst. Early diagnosis and treatment of heterotopic pregnancy in the presence of giant ovarian cyst is critical in improving favorable pregnancy outcomes.

**CASE PRESENTATION**

*Chief complaints*

No menstruation for more than 3 mo, vaginal bleeding for one day.

*History of present illness*

We are presenting a female 30-year-old, prim gravida, post assisted reproductive technology (IVF), 8w4d gestational age by scan, Chinese, Han tribe. She presented to our hospital with vaginal bleeding for one day. She had no history of headache, dizziness, abdominal pain, vaginal discharge, dysuria, diarrhea, coughing or fever.

*History of past illness*

History of past illness was non revealing.

*Personal and family history*

The patient had no reproductive history and denied any family history of hereditary diseases or malignancies.

*Physical examination*

The patient is conscious, mobile, cooperative in physical examination, and all vital signs are normal. Furthermore, physical examination was significant for blood on gloved finger on vaginal examination. The uterus was about the size of the second month of pregnancy. The bilateral adnexal area was thickened and there was no tenderness.
Laboratory examinations
The carbohydrate antigen 125 was 187 U/mL. Other routine blood and urine tests and other tumor markers showed no abnormalities.

Imaging examinations
B-Ultrasound on admission revealed an intrauterine pregnancy at 8w4d gestation age with heartbeat, enlarged left fallopian tube with a mixed mass of 3.6 X 3.4cm with active blood flow (Figure 1), and also a right ovarian mass of 4.9 X 4.3cm.

FURTHER DIAGNOSTIC WORK-UP
She was treated with magnesium sulfate and progesterone for fetal preservation. Reexamination after four days showed left fallopian tube with an uneven echo mass of 9.2 X 5.7cm and the right ovarian mass of 11.6 X 7.1cm. Close monitoring for the patient was paramount considering the abnormal tumor markers, rapidly growing ectopic pregnancy in case of spontaneous rupture and also the possibility of the ovarian mass of being malignant. These may affect the prognosis of the pregnant woman. After counselling the patient and relatives, we decided to perform laparoscopic exploration. Intraoperatively, the uterus was enlarged, left fallopian tube was enlarged by about 5 X 8 cm (Figure 2), right ovarian cystic mass of about 12 X 7cm was noted (Figure 3). The left ovary and right oviduct were normal. Patient underwent left salpingectomy for ectopic pregnancy removal and intrauterine pregnancy was preserved. A large theca lutein cyst of the right ovary was found during and resection was not done to avoid the decrease of progesterone which may lead to intra-abdominal pregnancy abortion. Pathology after left salpingectomy confirmed tubal pregnancy. One week after surgery, the patient developed severe pain in the right lower quadrant of which torsion of right ovarian cyst pedicle was not excluded. Instead of going for a second laparotomy for ovarian cystectomy, patient underwent right ovarian cyst fluid aspiration through the abdomen under ultrasound guidance and 300ml yellow liquid was aspirated. The patient's pain was subsequently relieved.
FINAL DIAGNOSIS
Combined with the patient’s medical history, physical examination and related examinations, the final medical diagnosis was left tubal pregnancy complicated by a giant theca lutein right ovarian cyst and intrauterine pregnancy.

TREATMENT
Three weeks after the operation, the patient was generally in good condition and recovered well, without complaints of discomfort, and was discharged. The gestation age on discharge was 12w3days, a viable fetus and reduced right ovarian cyst size.

OUTCOME AND FOLLOW-UP
At present, the pregnancy has delivered a baby successfully.

DISCUSSION
The clinical symptoms of an ectopic pregnancy are the same as those of an intrauterine pregnancy before rupture, and once the ectopic pregnancy ruptures, the presentation is similar to that of a simple ectopic pregnancy. However, in the absence of typical acute abdominal symptoms, the presentation may be limited to vaginal bleeding and mild abdominal pain, which is often misdiagnosed as preterm abortion and delays treatment. Serum HCG is one of the main tools for the diagnosis of ectopic pregnancy, but the time of multiplication of HCG in intrauterine pregnancy combined with ectopic pregnancy is not found to be significantly different from that of intrauterine pregnancy alone, especially after the second trimester, which lacks diagnostic significance. Ultrasound has some value in the diagnosis of this disease [3]. In the present case, a mixed echogenic mass was found outside the uterus, so ultrasound should not neglect the examination of the fetus in intrauterine pregnancy, especially in the bilateral adnexal area, to avoid the possibility of missing the combined ectopic pregnancy. The most common type of ovarian tumor found during pregnancy is benign teratomas, which is mostly detected before pregnancy and is seen as high-density signal on ultrasound. They are usually cystic or
solid with indistinct borders, often combined with a large amount of ascites and elevated CA125 and HE4, so the clinical situation needs to be fully evaluated to facilitate the diagnosis and differentiation of theca lutein ovarian cyst in pregnancy [8]. Theca lutein ovarian cysts are physiological ovarian cysts and hormone-related cysts, which can disappear spontaneously if the hormonal stimulation disappears and have the characteristic of self-generation as well as self-disappearance [5]. Therefore, surgery is not recommended for theca lutein ovarian cyst in pregnancy, but if the ovarian cysts are twisted or have severe bleeding causing ovarian rupture or shock, or if the ovarian enlargement causes obstructed labor, or Surgery is still recommended if ovarian tumors, especially malignant tumors are highly suspected [6, 7]. Therefore, it is recommended that patients with ovarian cysts in combination with pregnancy can be treated conservatively, but when a tumor is suspected, early detection and elective surgery should be performed to exclude potential malignant tumors or benign tumors with complications such as torsion of the ovarian cyst that could affect pregnancy outcome.

Once heterotopic pregnancy is diagnosed, the choice of treatment plan should be based on a combination of factors such as the number of weeks of pregnancy and whether the patient wants to keep the intrauterine pregnancy. For patients who wish to continue intrauterine pregnancy, laparoscopic technique is a safe and effective treatment method, which can replace traditional open surgery because of its advantages of early diagnosis and early treatment, less injury, faster recovery, fewer postoperative complications, and less interference with intrauterine pregnancy [8]. However, during laparoscopic salpingectomy or salpingostomy, there is inevitable uterine irritation that can lead to intrauterine miscarriage. Meanwhile, salpingotomy has the possibility of some products of conception remaining leading to persistent ectopic pregnancy. Simultaneously patients treated with salpingectomy also had a lower rate of clinical pregnancy than those treated with salpingostomy or those managed expectantly [9]. Patients and their families should be fully informed of these risks. In this case, the patient strongly requested for fetal preservation. In order to ensure the continuation of the intrauterine pregnancy and
complete removal of the extra uterine pregnancy, the operation time was minimized and stimulation to the uterus was minimal. To open the abdomen, we chose a low transverse incision close to the base of the uterus to reduce mechanical stimulation of the uterus while exposing the adnexa [10]. Intraoperative mechanical stimulation of the uterus and destruction of the corpus luteum of pregnancy should be minimized. Postoperative pain is also one of the causes of miscarriage, Therefore, postoperative analgesics can be given to pregnant women, as well as magnesium sulfate to inhibit contractions and progesterone in order to prevent miscarriage. Therefore, for heterotopic pregnancy complicated by a giant ovarian cyst, it is recommended individualized depending on the fertility requirements. If the intrauterine embryo is well developed, laparoscopic surgery is feasible to remove the affected fallopian tube and preserve the intrauterine embryo so that the intrauterine pregnancy can continue. Normal pregnancy with ovarian flavin cysts can be diagnosed based on their clinical manifestations and combined with ultrasound examination. If complications such as ovarian torsion do not occur, they can be observed until six months after the end of pregnancy, when most of them disappear spontaneously. If ovarian torsion occurs and surgery is required, conservative surgery can be performed to minimize damage to the ovaries and to continue the pregnancy before full term [11]. In this reported case, patients with theca lutein ovarian cyst with pedicle torsion cannot be ruled out by ultrasound but guided needle ovarian cyst aspiration can be done to relieve symptoms. The key to the treatment of this disease is early diagnosis, early treatment and correct differential diagnosis of various ovarian tumors to prevent ectopic pregnancy mass rupture, hemorrhage, or ovarian tumor torsion leading to tissue ischemia and necrosis, which can seriously endanger the life of the pregnant woman and intrauterine embryo, pay attention to reduce the damage during surgery, avoid affecting the blood circulation of the uterus and ovaries, avoid unnecessary surgical interventions after surgery, and also cooperate with drug therapy, which can improve The success rate of continuing pregnancy.
CONCLUSION

Approach in a patient with heterotopic pregnancy and giant ovarian cyst is individualized depending on the fertility requirements. We recommend that:

1. If the patient has satisfied parity (no fertility requirement), laparoscopic salpingectomy and the giant ovarian cyst and removal of intrauterine pregnancy.

2. If the patient has fertility requirement (willing to have more children in future), laparoscopic salpingectomy or salpingostomy can be done with the preservation of the intrauterine pregnancy. Serial ovarian cyst aspiration can be done under ultrasound; resection can be done after delivery.

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