Ovarian pregnancy: Two case reports

Jayeeta Roy¹, Anindita Sinha Babu²

1. Assistant Professor, Department of Obstetrics & Gynaecology, College of Medicine and JNM Hospital, WBUHS, Kalyani, India

2. Assistant Professor, Department of Pathology, College of Medicine and JNM Hospital, WBUHS, Kalyani, India

CASE REPORT

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Corresponding Author:

Jayeeta Roy, DGO, MS(OBGN) Assistant Professor Department of Obstetrics & Gynaecology College of Medicine and JNM Hospital, WBUHS, Kalyani, Nadia, India -741235, India. Email:<u>roymitrajayeeta@gmail.com</u>

Abstract

Ovarian pregnancy is a rare event occurring in 1-3% of all ectopic pregnancies. Increased reporting might be due to the wider use of intra-uterine devices, ovulatory drugs and assisted reproductive techniques. Though ovarian pregnancy has a distinct pathology, it can be a source of clinical and intraoperative diagnostic difficulty. We report two cases of ovarian pregnancy – one primary and one secondary – that came to our notice within six months span. Unlike tubal ectopic and secondary ovarian pregnancies, patients with primary ovarian pregnancy are likely to experience success in future intra-uterine conception and negligible risk.

Key Words

Ectopic pregnancy, ovarian pregnancy, assisted reproductive techniques, haemorrhagic ovarian cyst, corpus luteal cyst, pelvic inflammatory disease

Implications for Practice:

1. Ovarian pregnancy is a rare event constituting 1-3% of all ectopic pregnancies.

2. Ovarian pregnancy is increasing and primary ovarian pregnancy has better outcome than secondary ovarian or tubal ectopic pregnancy.

3. A high index of suspicion is required for diagnosis to avoid a crisis situation in the ward or operation theatre.

Background

Ovarian pregnancy is a rare event with incidence that estimates between 1 in 2,100 to 1 in 7,000 pregnancies, or 3% of all ectopic pregnancies.¹ The incidence has increased in the last 50 years² with causes attributable to better diagnostic modalities, increased use of intra-uterine devices, ovulatory drugs, and assisted reproductive techniques.³ A history of pelvic inflammatory disease (PID) has been implicated in the increase, as well.⁴

PID can result in a reduction of tubal motility⁵ and/or a thickening of the ovarian albuginea secondary to the inflammatory response. The latter will cause a reduction of follicular dehiscence and a subsequent increased risk of intra-follicular pregnancy. Moreover, ovarian pregnancy is frequently misdiagnosed clinically as a tubal ectopic pregnancy. Intraoperatively, it is difficult to differentiate among ovarian pregnancy, ruptured corpus luteal cyst, and haemorrhagic ovarian cyst. There is an obvious need to differentiate between tubal ectopic and ovarian ectopic pregnancies as well as between primary and secondary ovarian pregnancies. Thus, herein lies the necessity of reporting our two cases.

Case Report 1

A 22-year-old primigravida presented with lower abdominal pain, mild vaginal bleeding with nine weeks of amenorrhoea. She had a history of oligomenorrhoea. She did not have any past history of pelvic inflammatory disease or insertion of an intrauterine device. Tachycardia (140 bpm), blood pressure 90/60 mmHg, with abdominal



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guarding and tenderness were noted on examination. Per vaginal inspection revealed mild bleeding and bi-manual examination was slightly painful with the presence of an orange-sized right forniceal mass. Haemoglobin was 6.2gm/dl and urine was positive for beta hCG. Transvaginal sonography showed a gestational sac with a foetus in the region of the right ovary with a crown rump length of 16mm corresponding to the eighth gestational week, and with intra-abdominal haemorrhage. Culdocentesis was positive for haemoperitoneum. Ruptured ectopic pregnancy was diagnosed and emergency laparotomy was performed. The operative findings revealed ruptured right ovarian pregnancy. Unilateral salpingo-oophorectomy was performed on the right side and the specimen was sent for histopathological examination.

The ovary was enlarged (5.5 cm x 4 cm x 3 cm) with a foetus attached to its hilum via umbilical cord (Figure 1). On cross section of the ovary, a gestational sac with areas of haemorrhagic infarct was noted. Sections were taken for histopathological examination. The microsections showed multiple chorionic villi lined by trophoblastic cells along with deciduas. There was extensive haemorrhagic necrosis of the ovarian stroma (Figure 2). The sections from the fallopian tube showed no evidence of gestational tissue. Thus, a diagnosis of primary ovarian pregnancy was made as per Spiegelberg's criteria.⁵

The patient had an uneventful postoperative period and was discharged without complications. Her subsequent fertility was unaffected by the laparotomy; she presented with an intrauterine pregnancy three months later.

Figure 1: Ovary and part of fallopian tube with foetus attached to hilum via umbilical cord; ovary showing haemorrhagic infarct



Figure 2. Multiple chorionic villi lined by trophoblastic cells along with sheets of decidualised epithelium with a background of necrotic ovarian stroma (H&E stain, 100x magnification)



Case Report 2

A 22-year-old multigravida woman presented in shock with the history of amenorrhoea for six weeks, excruciating abdominal pain, history of syncope, severe pallor, with beta hCG level of 5,070.30 U/L and haemoglobin 4.7gm/dl. She had taken three cycles of medication for ovulation induction in the past, but had conceived spontaneously. There was no history of spontaneous or induced miscarriage, intrauterine device use, or pelvic inflammatory disease. Transvaginal sonography showed a normal uterus with an endometrial thickness of 17mm and a heterogenous echogenic mass on the right adnexa (8.7cm x 5.5cm x 5cm) with significant intraperitoneal bleeding. The left adnexae was normal. Per vaginal examination showed no vaginal bleeding and bimanual examination revealed fullness in the Pouch of Douglas (POD), a tender mass in the right fornix, and the presence of cervical excitation. Emergency exploratory laparotomy was performed. Operative findings revealed a ruptured right ovarian pregnancy with 1700mL of intraperitoneal bleeding. The left tube and ovary were normal. A tubo-ovarian mass was found on the right side consistent with the transvaginal sonogram. There was no evidence of endometriosis or chronic inflammation in the pelvis. A unilateral salpingo-oophorectomy was performed on the right side and the specimen was sent for histopathological confirmation. The patient received five units of grouped and cross-matched O-positive blood intraoperatively.

The patient's recovery was uneventful and she was discharged on the fifth day post-surgery with a beta hCG level of 1,430 U/L.



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The specimen of the right ovary with part of fallopian tube measuring 8cm x 5cm x 3cm with a tube 4cm in length and 2cm in diameter was received in the pathology department. The surface was congested. The cross-section of the ovary showed the ovarian tissue was replaced by black, friable necrotic tissue, but a gestational sac was identified (Figure 3). The cross-section of fallopian tube revealed blood clot. Histopathology sections from the ovary showed multiple chorionic villi lined by trophoblastic tissue with sheets of decidualised epithelium along with extensive haemorrhage and necrosis. No normal ovarian tissue was identified (Figure 4). The sections of the fallopian tube showed a normal lining of epithelium with multiple chorionic villi and deciua (Figure 5). A diagnosis of secondary ovarian pregnancy was made.

Figure 3: Cut section of ovary with a gestational sac and surrounding ovarian stroma showing haemorrhage and necrosis.



Figure 4. Microsection of ovary with multiple chorionic villi and extensive areas of haemorrhage (H&E stain, 100x magnification)



Figure 5: Microsection of fallopian tube with normal lining epithelium and multiple chorionic villi (H&E stain, 40x magnification)



Discussion

Ovarian pregnancies constitute three per cent of ectopic pregnancies.¹ Younger age, endometriosis, pelvic inflammatory disease, intra-uterine devices, use of ovulatory medications, and assisted reproductive techniques³ are a few risk factors that have contributed to the increased incidence of ovarian pregnancy. Since approximately 74 per cent of women with ovarian pregnancy are unaware,⁴ incidence is likely to be underestimated as many conceptuses die and involute spontaneously. More than 90 per cent of ovarian pregnancy cases present acutely. Moreover, some of the suspected tubal pregnancies that are treated conservatively with methotrexate, without laparoscopic validation, could be, in actuality, early ovarian pregnancies.⁶

Abdominal pain, with or without vaginal bleeding, is a common clinical symptom along with symptoms similar to those of tubal pregnancies, including circulatory collapse.⁶ In the first case-report, the patient denied any history of risk factors for ovarian pregnancy such as fertility therapies, pelvic inflammatory disease or intrauterine device insertion. It can be hypothesized that this primary ovarian pregnancy resulted from intrafollicular fertilisation that took place following failure of ovum extrusion after follicularrupture.

As for other ectopic locations, the combination of symptoms of acute abdomen with a history of antecedent amenorrhoea, raised beta hCG levels, and an ultrasonographically empty uterus should also trigger an investigation for not only tubal pregnancy, but also ovarian ectopic pregnancy.



Culdocentesis remains a valuable diagnostic tool. Its results facilitate decision-making to select an appropriate surgical approach: diagnostic laparoscopy or direct laparotomy. Laparotomy was performed in both of our cases, as the patients arrived in haemodynamically unstable condition. The established modes of surgical treatment of ovarian pregnancy are either removal of the entire ovary containing the ectopic gestation or performing a wedge resection of the ovary. It is often quoted that a wide wedge resection of the ovary might sometimes lead to a decrease in ovarian blood flow and function.⁶ In both the cases, after assessment of the lack of vitality of the ovarian tissue of the affected side, salpingo-ophorectomy was performed instead of wedge resection of the ovary. Medical treatment was not tried as the patients' condition demanded immediate surgery. However, etoposide and methotrexate use was contemplated after surgery if beta hCG levels remained elevated, indicative of persistent trophoblastic tissue.⁷

Over a century ago, Spiegelberg⁵ described criteria for an ovarian pregnancy. They were: the fallopian tubes, including fimbria, must be intact and separate from the ovary; the gestational sac must occupy the normal position of the ovary; the ovary must be attached to the uterus through the utero-ovarian ligament; and, there must be ovarian tissue attached to the pregnancy in the specimen (e.g. placental tissue has to be mixed with ovarian cortex).⁵ Histologically, the first case revealed placental and trophoblastic tissue in the ovarian tissue only, indicative of the ovarian origin of the ectopic pregnancy. In the second specimen, there was trophoblastic tissue in both the ovary and fallopian tube of the location of the ectopic pregnancy was made.

The differential diagnosis for ovarian pregnancy, both sonographically and at the time of surgery, remains a clinical challenge – that is to distinguish an ovarian ectopic pregnancy from a corpus luteum or haemorrhagic cyst⁸ or even ruptured chocolate cyst. A corpus luteum in an early or failing intrauterine or tubal pregnancy can mimic a cystic adnexal mass without clear intrauterine gestation.

During laparotomy in the second case, the adnexal mass resembled a hemorrhagic cyst in the right ovary. Macroscopically ovarian pregnancy can resemble an ovarian haematoma, clear ovum and embryonised ovum less than three months in size. Histology alone confirms the diagnosis as well differentiates between the different types of ovarian pregnancy.³

The patient with primary ovarian pregnancy subsequently conceived spontaneously in three months' time. This is

typically the outcome following a primary ovarian pregnancy as, unlike tubal pregnancy and secondary ovarian pregnancy, fertility is preserved and recurrence is a rarity.⁹

In conclusion, ovarian ectopic pregnancy is rare and expected to rise as more patients opt for fertility therapy. Despite modern diagnostic modalities, these patients continue to present in circulatory collapse. The necessity to maintain a high index of suspicion is required to ensure an efficient mode of treatment, appropriate prognosis, and patient counselling. Usually surgical treatment in form of oophorectomy or wedge resection of the ovary is required. Diagnosed properly, and in spite of the morbidities associated with ovarian pregnancy, a successful subsequent pregnancy can be expected following a primary ovarian pregnancy.

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CONFLICTS OF INTEREST

The authors declare that they have no competing interests.

ETHICS COMMITTEE APPROVAL

COMJNMH Institutional Ethics Committee

PATIENT CONSENT

The authors, *Jayeeta Roy and Anindita Sinha Babu*, declare that:

- **1.** They have obtained written, informed consent for the publication of the details relating to the patient(s) in this report.
- **2.** All possible steps have been taken to safeguard the identity of the patient(s).
- **3.** This submission is compliant with the requirements of local research ethics committees.