

Ovarian Pregnancy - a case report and literature review

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Abstract:

Ovarian pregnancy is one of the rarest types of ectopic pregnancies. Diagnosis is often challenging as it is confused with tubal pregnancy due to similar clinical features and imaging findings. Ovarian pregnancy is mostly diagnosed per operatively and the final diagnosis is only possible by histopathology. Because of late diagnosis, most of the patients present with rupture leading to catastrophic intra peritoneal hemorrhage and shock leading to removal of affected ovary. But some cases of ovarian pregnancy can be treated conservatively if there is an early diagnosis and patient is hemodynamically stable. So, high index of clinical suspicion and meticulous investigations are required to diagnose and manage this rare but dangerous condition. We hereby report unusual presentation and successful management of a case of ovarian pregnancy in our institute.

Key words: Pregnancy, ectopic pregnancy, ovarian pregnancy

Introduction:

Ectopic pregnancy is a condition where fertilized ovum is implanted in an area other than the normal uterine cavity. Among all the sites for ectopic implantation, ovary is quite a rare one. The incidence of ovarian pregnancy estimates to be between 1 in 2,100 to 1 in 7,000 pregnancies, or 3% of all ectopic pregnancies¹. The first case of ovarian pregnancy was first described in 1689 by De Saint Maurice of Perigod and the diagnosis was made postmortem². Once a rare finding, now there is a subtle rise in incidence of ovarian pregnancy in last few decades³. This rise may be subjective due to availability of better diagnostic facilities but also may be due to actual increase in risk factors like intrauterine contraceptive device, pelvic inflammatory disease and assisted reproductive technologies.⁴

Case Report:

A 36 years old regularly menstruating woman presented in outpatient consultation with 9 weeks of amenorrhea and a history for scanty per vaginal

bleeding and severe lower abdominal pain three weeks back. She went to a local physician where she was diagnosed as 9 weeks pregnancy. There was no history of spontaneous or induced miscarriage, intrauterine device use, or pelvic inflammatory disease. She did not receive any ovulation induction. Her serum β -hCG report came out as 2102 mIU/mL 4 days back, but the ultrasonography of lower abdomen on the same day showed a complex lesion having echogenic components and cystic area in right adnexa measuring about 7.2X4.4 cm. Mild fluid collection and high marginal vascularity was seen around the lesion.(Fig:1) Left ovary was normal. The report was consistent with right sided ectopic pregnancy. The patient had mild dull lower abdominal pain at the time of presentation but was hemodynamically stable. Her vital signs were - blood pressure 110/70 mmHg, pulse 96 beats per minute (bpm), temperature 36.5 °C. A physical examination showed minimal tenderness in all sides of her abdomen. She got admitted in Farida Clinic and Infertility Management Center on the next day. Her Hb level was 8.2 gm/dl. She had pre-operative

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blood transfusion for correction of anemia and underwent laparotomy. Upon opening the abdomen, uterus, left tube and ovary were found to be normal. On the right side, there was a pregnancy sac containing large amount of clotted blood with a gestational sac attached to the right ovary. Right tube was edematous but intact. (Fig: 2) There was large amount of clotted blood in the abdominal cavity which was removed. Right sided salpingo-oophorectomy was done and the specimen was sent for histopathology. As it was her 6th pregnancy with history of three term pregnancies with healthy children delivered by caesarian sections, one spontaneous abortion and one ectopic pregnancy, left sided tubal ligation was also done. One unit of blood was transfused per operatively. Her post-operative period was uneventful and she was discharged from the hospital with standard post-operative advice. She was advised for a follow-up visit after seven days with the histopathology report.

The sections from the fallopian tube showed no evidence of gestational tissue which confirmed our diagnosis of ovarian pregnancy.

Discussion:

Ovarian pregnancy accounts for approximately 1-3% of ectopic pregnancies. Ectopic pregnancy is more likely to happen if there is pelvic adhesion due to previous pelvic surgery, pelvic endometriosis or pelvic inflammatory disease. It is also common if there is an intrauterine contraceptive device in situ or history of ovulation induction and assisted reproductive technologies. Risk factors particularly associated with ovarian pregnancy are ill defined. The most important risk factor associated with ovarian pregnancy is supposed to be intra uterine contraceptive device in situ. Berger and Blechner in their study have shown that the ratio of ovarian ectopic pregnancy among women using an IUD to all ectopic cases is 1:9 as opposed to its prevalence in the general population which is detected as 1:150 to 200.⁶ In their study, Lehfeldt and his team also showed that IUDs prevent uterine implantation by 99.5 % and tubal implantation

by 95.5 %; but there is no preventive effect on ovarian implantation.⁷In addition to the above-mentioned risks, it is hypothesized that primary ovarian pregnancy can even result from intrafollicular fertilization that take place following failure of ovum extrusion after follicular rupture. In our case, the woman had developed ovarian pregnancy without any recognized risk factors associated with ectopic pregnancy.

Ovarian pregnancy may be primary or secondary depending on whether tube has been ruptured or not. Primary type occurs if the ovum is fertilized within the follicle and secondary type occurs if fertilization took place in the ampullary part of the tube and then implanted in the ovarian stroma after the rupture or abortion from ipsilateral tube. Primary ovarian pregnancy is quite rare and some even believe that there is no such thing as primary ovarian pregnancy and all the ovarian pregnancies diagnosed are actually a case of secondary ovarian pregnancy due to ruptured tubal pregnancy of that side.

Ovarian pregnancy can also be classified as acute and chronic. About 90% of ovarian pregnancies present as acute condition because it is usually terminated by a rupture in the first trimester. Because of the increased vascularization of the ovarian tissue, it leads to internal hemorrhage and hypovolemic shock. Some rare cases that reach second trimester are also documented. Stanley and Harris in 1994 reported a rare case of second trimester ovarian twin pregnancy.⁸ Most of the chronic ovarian pregnancies are often diagnosed as unruptured tubal pregnancy and managed conservatively without accurate diagnosis.⁹ Only proper histopathological evaluation can differentiate and ovarian from a tubal pregnancy.

Spiegelberg in 1878 first described the criteria for diagnosis of an ovarian pregnancy. These were:

- a. The fallopian tubes, including fimbria, must be intact and separate from the ovary; the gestational sac must occupy the normal position of the ovary
- b. The ovary must be attached to the uterus through the utero-ovarian ligament; and,

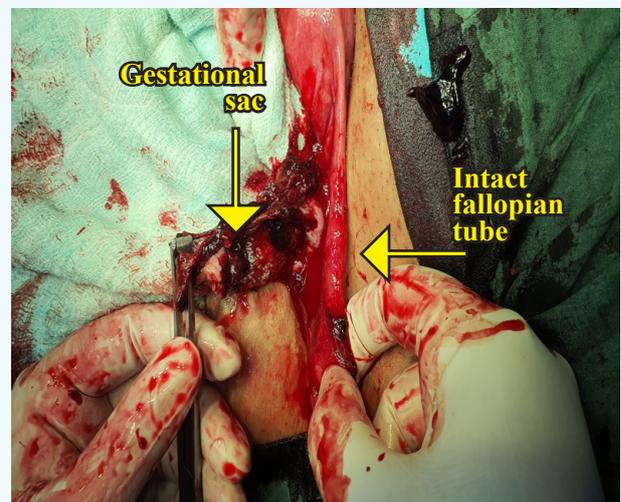
c. There must be ovarian tissue attached to the pregnancy in the specimen (e.g., placental tissue has to be mixed with ovarian cortex)¹⁰

Acute ovarian pregnancy is often clinically confused with other acute condition like ruptured corpus luteal cyst and ruptured tubal pregnancy. In all these conditions, patients present with a short period of amenorrhea, scanty or no per vaginal bleeding and lower abdominal pain. On ultrasound, there is an empty uterus with an adnexal mass adjacent to the ovary or, as has been described, a double echogenic ring within a hypo echoic latero-uterine mass.¹¹ In most cases, it is quite difficult to differentiate ovarian pregnancy from tubal pregnancy depending on clinical features and imaging techniques only. Hallat, in his study of 25 patients with primary ovarian pregnancy showed that a preoperative diagnosis was achieved in only in 28% cases, the rest were diagnosed by histopathological assessment postoperatively.¹² But there is marked improvement in ultrasonographic skills and instrumentation in recent years. Especially with the widespread use of the transvaginal scan, a bigger number of ovarian pregnancies can be diagnosed preoperatively. Ghi and Marconi in 2005 worked on 3Dscans to identify ovarian pregnancy

and showed that some criteria are very suggestive for sonographic diagnosis of ovarian pregnancy:

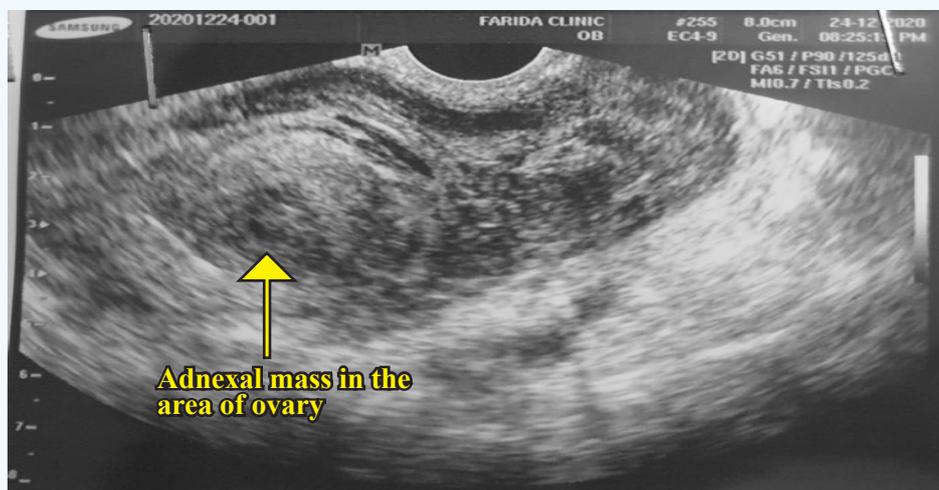
- a. An wide echogenic ring with an internal echolucent area on the ovarian surface
- b. The presence of ovarian cortex, including corpus luteum or follicles around the mass
- c. The echogenicity of the ring usually greater than that of the ovary itself.¹³

Fig. 2: Peroperative finding showing an adnexal mass attached to the ovarian ligament with intact fallopian tube of the same side.



The mainstay of management of ovarian pregnancy

Fig. 1: USG showing adnexal mass at the area of the ovary



is surgical. Either oophorectomy with or without the adjoining tube is the surgery of choice in late cases. Partial removal/ wedge resection of ovary may be attempted if fertility is desired but it may result in reduced ovarian vascularity and volume resulting in reduced ovarian reserve. Laparoscopy is usually preferred over laparotomy if the patient is hemodynamically stable.

Early diagnosis of ovarian pregnancy by serum β -hCG and an accurate transvaginal scan can keep the window open for more conservative approach like methotrexate, prostaglandins, potassium chloride, and hypertonic glucose administration¹⁴. These drugs halt trophoblast development and help preserve the normal anatomy crucial for future fertility preservation. Medical treatment with methotrexate is the most acceptable medical treatment now-a-days but still these cases are few and literature is sparse. Annunziata et al suggested that the criteria for successful methotrexate therapy were a gestation sac <30 mm, absent fetal cardiac activity, and less than 6 weeks of gestation.¹⁵

Recurrence of ovarian pregnancy is a very rare incidence. Only a single case has been described by Mehmood and Thomas in 1985 which showed the involvement of the opposite ovary.¹⁶

Conclusion:

Ovarian pregnancy, once a rare condition is on the rise due to multiple risk factors. Majority of these patients present with an acute emergency with circulatory collapse and shock. To avoid this catastrophe, a more vigilant approach should be taken in women suspicious of having ovarian pregnancy. Early diagnosis and timely management are needed to manage these cases and save women from associated morbidity or mortality. Continued attention to this problem in the future is important in order to allow clinicians to make informed decisions about this rare but dangerous clinical situation.

Consent:

Verbal consent was taken from the patient after ensuring anonymity to the patient.

Acknowledgments: None

Conflict of Interest: None

References:

1. Comstock C, Huston K, Lee W. The ultrasonographic appearance of ovarian ectopic pregnancies. *Obstet Gynecol.* 2005;105:42–45.
2. Lurie S. The history of the diagnosis and treatment of ectopic pregnancy: a medical adventure. *Eur J Obstet Gynecol Reprod Biol* 1992;43:1–7.
3. Gaudoin MR, Coulter KL, Robins AM, Verghese A, Harretty KP. Is the incidence of ovarian ectopic pregnancy increasing? *Eur J Obstet Gynecol Reprod Biol.* 1996;70:141–143
4. Das S, Kalyani R, Lakshmi V, Kumar MLH. Ovarian Pregnancy. *Indian J of Pathology and Microbiology.* 2008;51(1):57–62.
5. Grimes HG, Nosal RA, Gallagher JC. Ovarian pregnancy: a series of 24 cases. *Obstet Gynecol.* 1983;61:174
6. Berger B, Blechner JN. Ovarian pregnancy associated with copper-7 intrauterine device. *Obstetr Gynecol.* 1978;52:597–600.
7. Lehfeldt H, Tietze C, Gorstein F. Ovarian pregnancy and the intrauterine device. *Am J Obstet Gynecol.* 1970;108(7):1005–9.
8. Stanley JR, Harris AA, Gilbert CF, Lennon YA, Dellinger EH. Magnetic resonance imaging in evaluation of a second trimester ovarian twin pregnancy. *Obstet Gynecol.* 1994;84:648–51.
9. Raziell A, Schachter M, Mordechai E, Friedler S, Panski M, Ron-El R. Ovarian pregnancy—a 12-year experience of 19 cases in one institution.

- Eur J Obstet Gynecol Reprod Biol. 2004 May 10;114(1):92–6.
10. Spiegelberg O. Zur Casuistik der Ovarialschwangerschaft. Arch Gynecol. 1873;13:73–79
11. Sergent F, Mauger-Tinlot F, Gravier A, Verspyck E, Marpeau L. Grossesses ovariennes: re'évaluation des critè`res diagnostiques. J Gynecol Obstet Biol Reprod. 2002;31:741–746.
12. Hallat J. Primary ovarian pregnancy. A report of twenty-five cases. Am J Obstet Gynecol. 1982;143:50–60.
13. Ghi T, Banfi A, Marconi R, et al. Three-dimensional sonographic diagnosis of ovarian pregnancy. Ultrasound Obstet Gynecol. 2005;26:102–104.
14. J. L. Allison, M. Aubuchon, J. D. Leasure, and D. J. Schust, "Hyperosmolar glucose injection for the treatment of heterotopic ovarian pregnancy," *Obstetrics and gynecology*, vol. 120, no.2, pp. 449–452, 2012.
15. N. Annunziata, E. Malignino, and R. Zarcone, "Ovarian pregnancy treated with methotrexate," *Panminerva Med*, vol. 38, no. 3, p. pp, 1996.
16. Mehmood SA, Thomas JA. Primary ectopic ovarian pregnancy, report of three cases. J Postgrad Med. 1985;31:219.