



Ovarian Ectopic Pregnancies: Diagnostic Dilemma and Optimizing Outcomes through a Systematic Approach to Management

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Received Date: May 25, 2023

Published Date: June 10, 2023

Introduction

Ectopic pregnancy in which refers to the implantation of the fertilized ovum outside the normal endometrial cavity is a potentially life-threatening gynaecological emergency. It is one of the leading causes of maternal deaths during early pregnancy. Compared to the tubal ectopic pregnancy, an ovarian ectopic pregnancy is very rare, but may result in increased maternal morbidity and mortality due to difficulties with timely diagnosis. We describe a case of ovarian ectopic pregnancy, and dilemma in diagnosis. We have recommended an acronym "OVARY" to ensure a timely diagnosis.

Case Review

A 31 year old, unmarried woman presented to the urgent care centre with an acute abdominal pain of 3 hours. Her menstrual history suggested regular cycles and last menstrual date was 6 days before presentation. Her past medical and surgical history was uneventful. On clinical examination, patient was hemodynamically stable & had a generalised abdominal tenderness.

She was investigated in the emergency department and all her routine blood tests were normal.

As she was unmarried, due to local cultural expectations, a serum B-hCG was not initially requested, and transvaginal ultrasound scan was not performed. An abdominal ultrasound & computed tomography (CT) scans showed a collection of fluid ?? ascities in the pelvis (Figure 1) and hepatorenal angle and echogenic focus at the right ovary with suspicion of cyst probably dermoid & rest abdominal and pelvic organs were reported as normal.

Initial diagnosis was an ovarian cyst rupture with with haemoperitoneum due to the acute abdominal pain or ascities secondary to an ovarian malignancy.

The woman's condition rapidly deteriorated and she appeared pale and became tachycardic & hypotensive. A serum B-hCG was requested and the results were suggestive of an ectopic pregnancy (3199 mIU/ml with an absent intrauterine sac).

An urgent laparoscopic surgery was carried out, which confirmed haemoperitoneum (Figure 2) with evidence of "cyst rupture" on the right ovary. Both Fallopian tubes were morphologically normal. In view of the rare possibility of an ovarian ectopic pregnancy, a biopsy was taken at the site of cyst rupture and the specimen was sent for histopathological examination. The patient made a good postoperative recovery. Histopathological examination confirmed an ovarian ectopic pregnancy.



Figure 1. Abdominal ultrasound scan showing a normal left ovary, with haemoperitoneum and a blood clot surrounding the right ovary.



Figure 2. Haemoperitoneum observed during the laparoscopy

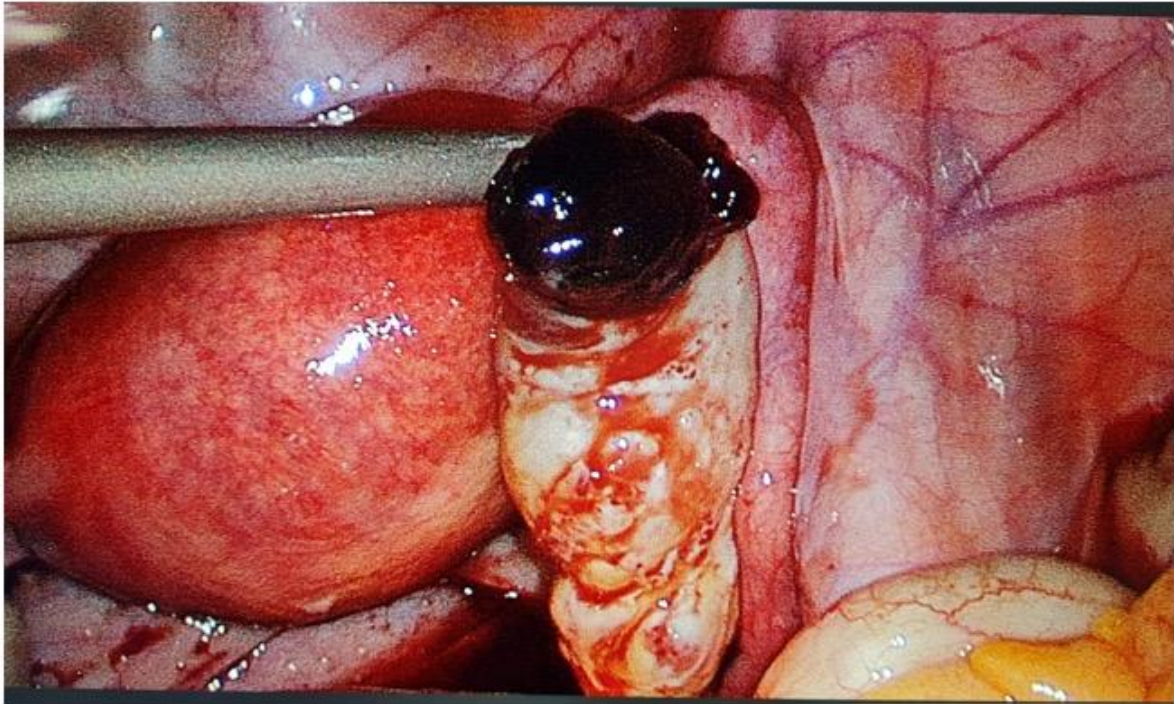


Figure 3. Site of rupture of the right ovarian ectopic pregnancy

<u>O</u>verall Clinical picture	Period of amenorrhoea, iliac fossa pain, history of IUD insertion
<u>V</u>aginal examination	Cervical excitation, tenderness in the fornices
<u>A</u>dditional pointers	Positive pregnancy test, absence of an intrauterine pregnancy on ultrasound with sonographic features suggestive of an ovarian ectopic pregnancy
<u>R</u>ecognition (intra-operative)	Normal Fallopian tubes, cyst / evidence of a ruptured cyst in one of the ovaries
<u>Y</u>ardstick	Histopathological confirmation of implanted trophoblasts within the ovarian substance and meeting all the 4 Spiegelberg's Criteria

Table 1. The Acronym "OVARY"

Discussion

We describe a case of a ruptured primary ovarian (ectopic) pregnancy, which is a very rare, potentially life-threatening complication during early pregnancy. It accounts for approximately 0.5–3.0% of all ectopic pregnancies, with an incidence ranging from 1 in 7000 to 1 in 2100 pregnancies.

Primary ovarian pregnancy occurs when the trophoblasts invade the ovary leading to the implantation of the gestational sac within the ovary and on the corpus luteum, and this usually results in the rupture of the ovary and a massive hemoperitoneum, Ovarian (ectopic) pregnancy, based on the location within the ovary, may be intrafollicular or extrafollicular. Intrafollicular ovarian ectopic pregnancy is mostly primary where fertilization occurs within the ovary. In contrast, an extrafollicular ovarian pregnancy can be either primary or secondary (1), in the latter, the fertilization occurs within the Fallopian Tube and the gestational sac subsequently gets implanted in the ovary.

The history of recent use of an intrauterine device (IUD) with extra uterine pregnancy should be viewed with a high index of clinical suspicion of an ovarian pregnancy because Razeiel et al., reported that 90% of ovarian pregnancy occurred in IUD users (6). Lehfeltdt et al, concluded that IUD prevents intrauterine implantation in 99.5%, tubal implantation in 95% and none at all in ovary (7). However, in our case we had no such history of previous IUD use.

Diagnostic Challenges

The pre-operative diagnosis of an ovarian pregnancy is often very challenging. The clinical presentation is similar to those of tubal pregnancy wherein both may have amenorrhea, irregular vaginal bleeding, abdominal pain and adnexal mass. An ovarian ectopic pregnancy is often misdiagnosed both clinically (iliac fossa pain, same as a tubal ectopic pregnancy) and sonologically as a ruptured tubal ectopic pregnancy, a ruptured corpus luteum cyst, a haemorrhagic cyst or an endometriotic (chocolate) cyst of ovary. In the Middle East, due to societal norms and expectations, the woman did not disclose her period of amenorrhoea, and as per our routine clinical practice, serum B-hCG was not requested as the woman was unmarried to respect the cultural sensitivity.

It is often very difficult to differentiate an ovarian pregnancy from a haemorrhagic ovarian cyst at the time of surgery (Figure 3). Due to the rarity of this condition, our intra-operative diagnosis was a ruptured haemorrhagic cyst. However, due to the positive pregnancy test, we took a sample of the ovarian tissue at the site of rupture, which showed implanted trophoblasts, clinching the diagnosis of an ovarian ectopic pregnancy.

Hallet et al (1982) studied 25 cases of ovarian pregnancies and reported that a correct surgical diagnosis was only made in 28% of cases. In the remaining cases, it was the pathologist who made the diagnosis [2]. It is important to appreciate that an ovarian pregnancy can terminate several days before the expected date of menstruation without any history of amenorrhea. Rimdusit et al reported 10 cases of ovarian pregnancy, out of which six had no history of amenorrhea (3). Their findings are similar to our case.

If a woman with an ovarian ectopic pregnancy presents before its rupture, then, it is possible to make a pre-operative diagnosis with a high-resolution transvaginal ultrasonography, with colour Doppler, confirming a gestationa sac / embryo with a fetal heart beat on the ovary. Ultrasound features of an ovarian pregnancy include.

- Wide echogenic ring with an internal echo lucent area on the ovarian surface. The echogenicity of the ring is usually greater than that of the ovary itself, compared to a thin tubal ring with tubal pregnancies or corpus luteum cyst.
- A yolk sac or embryo was less commonly seen as progression beyond early stages is exceptional [4].
- Other sonographic findings like complex adnexal masses or solid cystic masses with or without fluid in cul de sac, fluid surrounding the ovary and ovarian enlargement also result in high suspicion towards ovarian pregnancy [5].

In the presence of ultrasound findings strongly suggestive of an ovarian ectopic pregnancy, a diagnostic laparoscopy will also confirm normal Fallopian Tubes with an unruptured gestational sac implanted on the surface of the ovary. A very high index of clinical suspicion is mandatory in the presence of a positive β HCG without an intrauterine gestation or evidence of tubal ectopic pregnancy, and presence of a cystic structure on the surface of the ovary. In such situations, a sample of ovarian tissue adjacent to the “cystic structure” should be taken for histopathological examination to confirm or to exclude the diagnosis of an ovarian ectopic pregnancy.

Management options

Our pre-operative and intra-operative diagnosis, based on the clinical and imaging information available to us for a ruptured tubal ectopic pregnancy. Therefore, we proceeded with an emergency laparoscopy with an intention to perform a salpingectomy. In the presence of haemoperitoneum and absence of any evidence of a tubal ectopic pregnancy, and the presdence of a haemorrhagic area within

the ovary, our intra-operative diagnosis was a ruptured haemorrhagic cyst. However, in the presence of a positive pregnancy test, we took a biopsy to exclude an ovarian ectopic pregnancy.

Early diagnosis and treatment of an ovarian ectopic pregnancy is absolutely necessary to ensure a successful outcome. Seinere et al., concluded that laparoscopy is required for diagnosis and also for effective definitive surgical management at the same time [8].

In an unruptured tubal pregnancy, if certain criteria are met, the use of MTX is now a well-established and, in carefully selected cases, conservative treatment with serial estimation of B-hCG may be carried out (13). However, in contrast to a tubal ectopic pregnancy, medical treatment by methotrexate does not have robust scientific evidence to be considered as a safe and an effective treatment option for an ovarian ectopic pregnancy. Therefore, it is not recommended in routine gynaecological practice. Kudo et al. were the first to successfully treat an ovarian pregnancy with MTX, followed by Shamma et al. who used a single- intramuscular dose of MTX (50 mg/m²) (15). Mittal was the first to report an MTX injection directly in the gestational sac (16). Spontaneous bleeding after the rupture of ovarian pregnancy is the main complication, and high mortality rates have been reported with mortality (12)

Therefore, surgical treatment is recommended for an ovarian ectopic pregnancy and conservative surgical management such as ovarian wedge resection may be attempted depending on the extent of tissue destruction. Oophorectomy is not recommended as a routine management and preservation of ovary should be always given precedence [9]. This is because the patients are generally fertile and the risk of recurrence is zero as no case of recurrent ovarian pregnancy has ever been documented. This is in contrast to tubal pregnancy which has approximately 15% recurrence [10]

The definitive diagnosis of an ovarian ectopic pregnancy should be made according to the Spiegelberg's Criteria [11].

The following 4 criteria should be satisfied:

1. The tube on the affected side should be intact separate from the ovary.
2. The gestational sac must be connected to the uterus by the utero-ovarian ligment.
3. The gestational sac should occupy the normal position of the ovary.
4. Definite ovarian tissue must be present in the gestational sac wall.

In our case, we did ovarian conservation surgery with excision of ectopic trophoblastic tissue, and haemostasis was achieved with bipolar coagulation. The ectopic tissue was removed with an endobag through 10 mm trocar and the specimen was sent for a histopathological examination, which confirmed

the diagnosis of an ovarian ectopic pregnancy. Both the Fallopian tubes were healthy and there were no signs of tubal miscarriage.

We made a definitive diagnosis of an ovarian ectopic pregnancy, retrospectively, based on the intra-operative findings, the declining B-hCG levels following surgery, histopathological confirmation of presence of chorionic villi and trophoblastic tissue within the ovarian tissue.

We have proposed the following acronym “OVARY” to help clinicians timely diagnose and appropriately manage an ovarian ectopic pregnancy, which is a rare, but a potentially life-threatening early pregnancy complication.

References

- 1- Check JH, Chase JS. Ovarian pregnancy with contralateral corpus luteum. *Am J Obstet Gynecol.* 1986;54:155–56. [PubMed] [Google Scholar]
- 2- Hallett JG. Primary ovarian pregnancy: A report of twenty-five cases. *Am J Obstet Gynecol.* 1982;143:50–60. [Google Scholar]
- 3- [4] Rimdusit P, Kastri N. Primary ovarian pregnancy and the intra uterine contraceptive device. *Obstet Gynecol.* 1976;48:57–59. [PubMed] [Google Scholar]
- 4- Comstock C, Huston K, Lee W. The ultrasonographic appearance of ovarian ectopic pregnancies. *Obstet Gynecol.* 2005;105(1):42–45. [PubMed] [Google Scholar]
- 5- [8] Ghi T, Banfi A, Marconi R, Iaco PD, Pillu G, Aloysio DD, Pelusi G. Three dimensional sonographic diagnosis of ovarian pregnancy. *Ultrasound Obstet Gynecol.* 2005;26:102–04. [PubMed] [Google Scholar]
- 6- Razeiel A, Golan A, Pansky M, Ronel R, Bukovsky J, Capsi E. Ovarian pregnancy: a report of twenty cases in an institution. *Am J Obstet Gynecol.* 1990;163:1182–85. [PubMed] [Google Scholar] [Ref list]
- 7- Lehfelddt H, Tietze C, Gorstein F. Ovarian pregnancy and intrauterine device. *Am J Obstet Gynecol.* 1970;108:1005–09. [PubMed] [Google Scholar]
- 8- Seiner P, Di Gregorio A, Arisio R, Decko A, Crane F. Ovarian pregnancy and operative laparoscopy report of eight cases. *Hum Reprod.* 1997;12(3):608–10. [PubMed] [Google Scholar]
- 9- Panda S, Darlong LM, Singh S, Borah T. Case report of a primary ovarian pregnancy in a primigravida. *J Hum Reprod Sci.* 2009;2:90–92. [PMC free article] [PubMed] [Google Scholar]

- 10- Sergent F, Mauger-Tinlot F, Gravier A, Versyck E, Marpeau L. Ovarian pregnancies: reevaluation of diagnostic criteria. *J Gynecol Obstet Biol Reprod.* 2002;31:741–46. [PubMed] [Google Scholar]
- 11- Spiegelberg O. Zur casuistic der Ovarialschwangerschaft. *Arch Gynaekol.* 1878;13:73–79. [Google Scholar]
- 12- Ovarian ectopic pregnancy. Jha S, Bosworth K, Quadri A, Ibrahim A. *BMJ Case Rep.* 2011;2011 [PMC free article] [PubMed] [Google Scholar] [Ref list]
- 13- Stovall TG, Ling FW, Buster JE. Outpatient chemotherapy of unruptured ectopic pregnancy. *Fertil. Steril.* 1989;51:535–538. [PubMed] [Google Scholar]
- 14- Kudo M, Tanaka T, Fuzimoto S. A successful treatment of left ovarian pregnancy with methotrexate. *Nippon Sanka Fujinka Gakkai Zasshi.* 1988;40:811–813. [PubMed] [Google Scholar]
- 15- Shamma FN, Schwartz LS. Primary ovarian pregnancy successfully treated with methotrexate. *Am. J. Obstet. Gynecol.* 1992;167:1107–1308. [PubMed] [Google Scholar]
- 16- Mittal S, Dadhwal V, Baurasi P. Successful medical management of ovarian pregnancy. *Int. J. Gynecol. Obstet.* 2003;80:309–310. [PubMed] [Google Scholar]