



Case Study

Ruptured ovarian ectopic pregnancy: Case Report and review of literature

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A B S T R A C T

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Ovarian ectopic is a rare and dangerous form of ectopic pregnancy. A little is known about its risk factors and incidence. We present a case of ruptured primary ovarian pregnancy which was a surprise intra operative finding. As assisted reproductive technology (ART) procedures are becoming popular the incidence is likely to increase. Clinicians should be well equipped to diagnose and treat this unusual form of ectopic pregnancy at the earliest.

Introduction

Ectopic pregnancy is one of the most common gynaecological emergencies. Ectopic pregnancy occurs in 2 % of all pregnancies, the myriad of locations have a hierarchy of prevalence as well as associated mortality and morbidity. The condition occurs once in every 25000–40000 pregnancies (Salas Valien *et al.*, 1995). The most common location of the ectopic pregnancy continues to be fallopian tubes (accounting for roughly 95% of all ectopic pregnancies) followed by far less common ovarian ectopic which comprises 0.3–3% of extra uterine cavity pregnancies (Hertig, 1951).

Ovarian pregnancy is a rare diagnosis of exclusion. ART procedures have resulted in a rise of pregnancy outside the uterine cavity but ovarian pregnancy is still rare. Primary ovarian pregnancy results when an ovum not released from the ovary is fertilized or following a primary implantation of

fertilized ovum in the ovary after reverse migration from fallopian tube. In secondary ovarian pregnancy there is a tubal abortion or rupture with secondary implantation of gestational sac on the surface of ovary. The first case was reported by St Maurice in 1689. The diagnosis can be established sonographically by a hyper dense chorial ring which moves with the movement of ovary (Stucki and Buss, 2008). Even with modern 3D ultrasound probes the diagnosis is difficult as the lesion is heterogeneous with mixed echoes and it mimics corpus luteum or a hemorrhagic cyst of ovary. Often, the ultrasound diagnosis with raised Beta human chorionic gonodotropin to our dismay is “pregnancy of unknown location”. One third of all pregnancies irrespective of location are self abortive. It is likely that the incidence of ovarian pregnancy is more than

recorded. Primary ovarian pregnancy can be classified as intrafollicular or extrafollicular (Kaur *et al.*, 2011). In intrafollicular pregnancy the etiology is hormonal resulting in a trapped ovum inside the follicle. Another postulated cause is thickened tunica albuginea of ovary. It may also result due to inefficient sweeping of fimbria ovarica across the surface of ovary resulting in ineffective ovum pick up (Zaidi *et al.*, 2012).

Case Presentation

A 29 year old lady presented to the emergency department with the complaints of sudden onset severe pain abdomen for 2 hours. Her last menstrual period was fourteen days back. She was parity 2, with two spontaneous vaginal deliveries. She had previous two spontaneous abortions, one at three months and the other at five months gestation. She was breast feeding her one year old child. Her previous menstrual cycles were regular with 3-4 days of bleeding in every 28–30 days. She was married for eight years. She had no history of contraceptive use. She gave no history suggestive of intake of ovulation induction drugs. She was hemodynamically unstable. There was tachycardia and hypotension. Lower abdomen tenderness, rigidity and guarding were present. Speculum examination was normal. Vaginal examination revealed tenderness in bilateral fornices. Her urinary beta human chorionic gonadotropin was positive.

Pelvic ultrasonography revealed empty uterine cavity and haemoperitoneum and a left adnexal mass of 7 × 6 × 6 cm. Pelvic CT scan did not reveal the location of pregnancy. An emergency laparotomy was done with a preoperative provisional diagnosis of a ruptured left tubal ectopic pregnancy. Intraoperatively, the uterus was normal in size and both tubes were normal. A ruptured left ovarian ectopic was found (Fig. 1). Abdomen was filled with 1500 ml of fresh blood clots. Right ovary was normal. Left ovary was enlarged and showed a breach on the surface with an active bleeder on ruptured surface. Left salpingo-ophorectomy was carried out as the fresh bleeding vessels on the ovarian surface could not be secured after plication and electrocauterization. Left ovarian artery was ligated after visualizing the ureteric course. Right partial salpingectomy was done using modified pomeroys technique as per the patient's desire. Haemostasis was secured. The patient had an uneventful post operative course. Histopathological study of the excised specimen showed trophoblastic tissue in scan view (Fig. 2, 3) and high power (Fig. 4). Postoperatively the diagnosis of ruptured primary ovarian ectopic pregnancy was made according to Spiegelberg's criteria (Gavrilova-Jordan *et al.*, 2006). As suggested by Spiegelberg (1878), primary ovarian pregnancy should be differentiated from distal tubal pregnancy, a condition that secondarily involves the ovaries after tubal rupture or abortion. It was outlined four points.

1. The fallopian tube with its fimbriae should be intact and separate from ovary.
2. The gestational sac should occupy the normal position of the ovary.
3. The gestational sac should be connected to the uterus by utero-ovarian ligament.
4. The ovarian tissue must be preserved in the specimen attached to the gestational sac.

Two units of cross matched and typed packed red blood cells were transfused in the immediate postoperative period. She was discharged after normalization of serum HCG levels. The patient followed after three months was asymptomatic, with regular menstrual cycles.

Figure 1(a). Microscopic histopathology of trophoblast in the shelled out gestational sac from the ovary

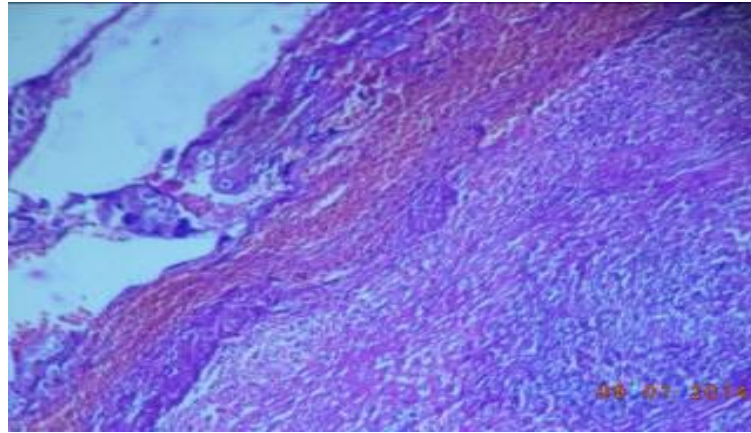


Figure 1(b). Microscopic histopathology of trophoblast in the shelled out gestational sac from the ovary

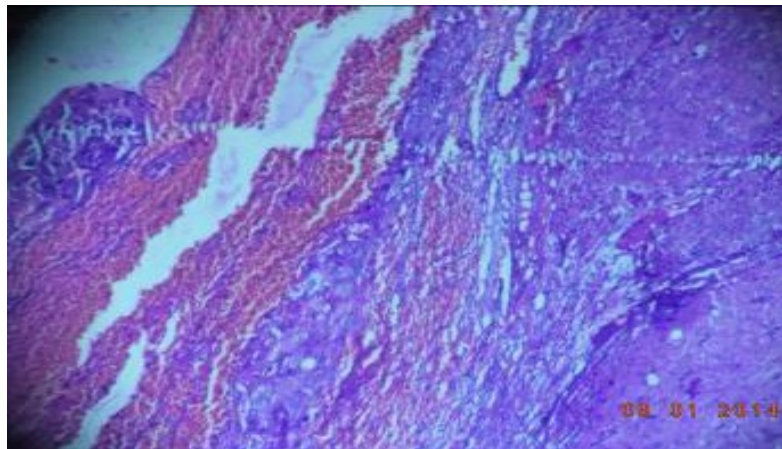
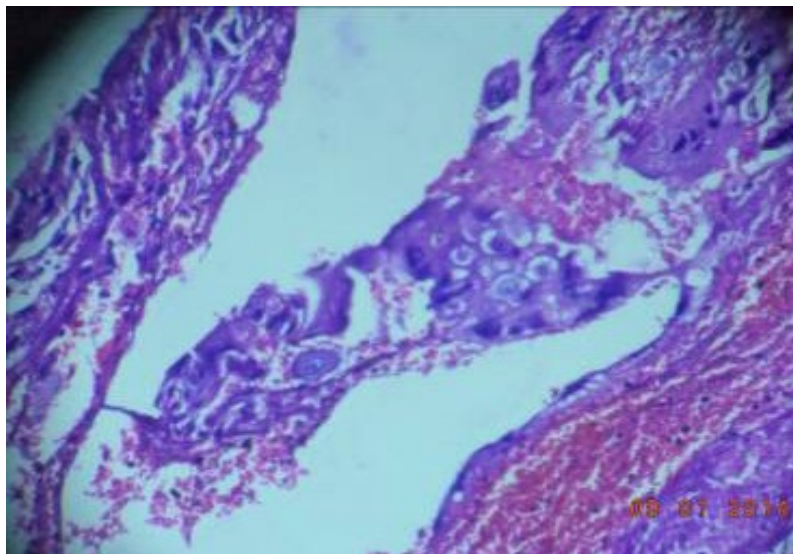


Figure 2. High power view of a trophoblastic cell (centre)



Discussion

There is often a delay in the diagnosis of ovarian pregnancy as the gestational sac mimics corpus luteum, hemorrhagic cyst and endometriotic cyst of ovary. In our patient, the diagnosis of interstitial ectopic pregnancy was missed on Ultrasonography and pelvic CT scan. It was a surprise intra operative finding. They pose a significant diagnostic and therapeutic challenge and carry a greater maternal mortality risk than tubal ampullary ectopic pregnancy. The developing chorionic villi may eventually erode into the blood vessels of ovary, causing a severe hemorrhage. Ovarian pregnancy occurs at the most richly vascularised site of female pelvis, junction of branches of uterine and ovarian arteries. Significant maternal hemorrhage leading to hypovolemia and shock can rapidly result from ovarian pregnancy rupture (Hertig, 1951). Clinicians should be aware of the difficulties with clinical, radiological and intraoperative diagnosis.

Little is known about the incidence and risk factors. The etiological factors for the ovarian pregnancy are pelvic inflammatory disease, fibroids, previous pelvic surgery and use of ART procedures (Spiegelberg, 1878). Altered tubal motility as a result of increased progesterone secretion from multiple corpora lutea or ovarian hypervascularity following hyperstimulation could be a cause (Gavrilova-Jordan *et al.*, 2006). A high number of transferred embryos, a transfer near the uterine horn, and excessive pressure on the syringe during the transfer or difficulties during the embryo transfer procedure increase the risk (Gavrilova-Jordan *et al.*, 2006). Peri and intra tubal adhesions related or not related to endometriosis are an additional risk factor. Some studies associate smoking as a risk as it interferes with tubal motility and ovum

pick up by fimbria ovarica (Stucki and Buss, 2008). Certain authors also consider the quality of embryos and the hormonal milieu at the moment of transfer as a possible cause (Kaur *et al.*, 2011). Interestingly the most common reported site is left ovary. This leaves the question of reverse migration of embryo open (Gavrilova-Jordan *et al.*, 2006).

With increasing use of ART procedures clinicians should be equipped with skills to diagnose this evasive form of ectopic as early as possible. The diagnosis of ovarian pregnancy is made by critical evaluation of all criteria used for other types of ectopic pregnancy. Symptoms are acute abdominal pain, intraperitoneal bleeding, a low hematocrit, and a positive serum or urine pregnancy test. Diagnostic tests include the sensitive beta-HCG radio immunoassay, culdocentesis and ultrasonography. Ovarian pregnancy is diagnosed with ultrasonographic criteria in presence of a positive HCG level indicating pregnancy.

The transvaginal ultrasound criteria for ovarian pregnancy are more echogenic white ring in the ovary compared with ovarian tissue. A yolk sac or fetal parts may be visualized but an embryonic pole is rarely seen (Kaur *et al.*, 2011). The corpus luteum has an anechoic texture and less wall echogenicity as compared to endometrium. Transvaginal three dimensional ultrasound scanning can reproduce the coronal plane of the uterus, facilitating exact localization of the gestational sac relative to uterine tube and ovary.

Features that are helpful with the use of 3-dimensional Trans Vaginal Ultrasonography (TVS) include a live embryo in a gestational sac surrounded by ovarian tissue. Another diagnostic aid is Laparoscopy, which has the advantage of allowing both diagnosis and

treatment. One study has shown that early diagnosis of ovarian pregnancy with TVS allows for first trimester conservative management with methotrexate (Chelmow *et al.*, 1994). No case of recurrent ovarian ectopic has been reported as contrast to 15% risk of recurrence of tubal ectopic pregnancy (9). However, if the diagnosis is made later in the gestation, as in our patient, surgical treatment with ovarian resection or oophorectomy may be required. The Royal College of obstetrician and gynecologists recommends that women with ectopic pregnancy most suitable for methotrexate therapy are those with serum beta HCG levels of 3000IU/ml and with minimum symptoms (RCOG, 2001).

Conclusion

Ovarian pregnancies are rare and can be missed radiologically and intraoperatively. Historically the obstetricians could diagnose them only after histopathological confirmation following excision according to Spiegelberg's criteria. With a rise in ART procedures the incidence is expected to rise. A laparoscopic visualization and frozen section biopsy of a gestational sac excised from the ovary can be used to diagnose ovarian ectopic in modern gynaecology

Learning points

- Ovarian pregnancies are rare and can be missed radiologically. The obstetrician should suspect a ruptured ovarian pregnancy when bilateral tubes are normal and the ovarian surface is breeched with haemoperitoneum.
- Historically the difficulties associated with early diagnosis of ovarian pregnancy are both radiological and intra operative. The gestational sac in the ovary is difficult to differentiate from corpus luteum or hemorrhagic cyst of

ovary. There is a role of three dimensional ultrasound to improve the detection rates.

- There is a role of frozen section biopsy to differentiate a ruptured corpus luteum from a ruptured ovarian pregnancy intraoperatively.
- Assisted reproductive procedures are on a rise. Clinicians should be well aware to diagnose the ovarian ectopic at the earliest.
- In our patient the ovarian gestation had ruptured and the only option was emergency salpingo-oophorectomy.
- Till date no case of recurrent ovarian ectopic has been reported. The exact risk factors and etiopathogenesis of ovarian ectopic are as yet unidentified.

Consent

We have obtained the patient's consent for the case report.

Competing interests

We do not have any commercial association that might pose a conflict of interest in connection with the manuscript. We certify that neither this manuscript nor one with substantially similar content under our authorship has been published or is being considered for publication elsewhere.

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