Case Report
Received on 16.06.2016, **Accepted on** 25.06.2016

Indian Journal of Pathology: Research and Practice
Volume 5 Number 3, September - December 2016
DOI: http://dx.doi.org/10.21088/ijprp.2278.148X.5316.28

A Clustering of Three Cases of Primary Ectopic Ovarian Pregnancy in a Rural Setting: Case Report and Review of Literature

Aditi Raina*, Shalini Trivedi, Malay Bajpai*, Indubhushan*****

*Associate Professor, ***Professor & Head, Department of Pathology, Rama Medical College, Ghaziabad. **Specialist Pathology, IGESI Hospital, Delhi.

Abstract

Primary ovarian pregnancy is one of the rarest types of ectopic pregnancies. Patients may complain of abdominal pain and bleeding per vaginum. Likely risk factors are intrauterine devices and assisted reproductive technology (ART). A pre-operative diagnosis is challenging but transvaginal sonography may be helpful. A diagnostic delay may lead to rupture, secondary implantation or operative difficulties. Therefore, awareness of this rare condition is important in reducing the associated risks. Here, we report three cases of primary ovarian pregnancies presenting with acute abdominal pain. Transabdominal ultrasonography could not be done in one, while in the second TVS could not particularly pin-point the site of the ectopic pregnancy. Both cases were confirmed by histopathological examinations.

Keywords: Primary; Ovarian; Pregnancy.

Introduction

Primary ovarian pregnancy is a rare condition accounting for 0.15% of all pregnancies and 0.15–3% of all ectopic gestations [1]. Unlike tubal ectopic, an ovarian ectopic pregnancy can present with mild symptoms and a delayed presentation which could be missed, posing a big diagnostic challenge. In the absence of well documented risk factors, a high index of suspicion is necessary to make a diagnosis.

The diagnosis of an ovarian ectopic pregnancy is seldom made before surgery. Ultrasound, especially transvaginal scanning (TVS) has proved to be an invaluable tool in the diagnosis of this condition [2].

Case 1:

A 26 years old female, with 45 days' amenorrhoea was admitted in the surgical emergency ward on 13.03.2014, which was followed by 2 episodes of bleeding PV at 13 day interval. She complained of pain right lower abdomen. The patient, was a fourth gravida, with a history of three full term normal vaginal pregnancies. On per vaginum examination, cervix and vagina were healthy and the uterus was enlarged to 7 weeks' size. The right fornix was thickened and tender.

Her routine hematological and biochemical tests were within normal limits except for mild leucocytosis with neutrophilia. The right tube appeared normal but the right ovary was enlarged and haemorrhagic. Right sided salpingo-oophorectomy with left sided tubal ligation was done and specimen was sent for histopathological examination.

On Gross examination, the ovary measured 4x3.5x2 cms; the cut surface showed blood clots and corpus luteum; no embryo was identified grossly. The fallopian tube, measuring 4cm, appeared normal. On microscopic examination, the sections showed corpus luteum within ovarian stroma along with extensive areas of haemorrhage and scattered chorionic villi.

Case 2:

A 38 years old female patient with 45 days amenorrhoea was admitted in the surgical emergency ward on 03.12.2014 with a history of pain in the right renal angle. There was no history of difficulty in

Corresponding Author: Aditi Raina, Associate Professor, Department of Pathology, Rama Medical College, Ghaziabad, Gaziabad – 245304, Uttar Pradesh
E-mail: rainaaditi@gmail.com

micturition, defaecation or vaginal bleeding.

On clinical examination she was pale; with a pulse rate of 98/minute and blood pressure of 120/80 mm Hg. The abdomen appeared slightly distended with tenderness in both iliac fossae. The vaginal examination revealed tenderness in all the fornices. A clinical diagnosis of a possible ruptured ectopic pregnancy or an acute PID was made.

USG done showed heterogenous solid mass at the right adnexa lesion with mild free fluid in the peritoneum suggestive of ruptured right tubal ectopic. On laparotomy, hemoperitoneum was seen and about 1 litre of blood was evacuated. Right sided salpingo-oophorectomy with left sided tubal ligation was done and specimen was sent for histopathological examination. The patient was admitted in MICU and 2 units of PRBCs were transfused. The patient was discharged on 5-12-14 in stable condition. Hb-10.3gm, TLC was mildly raised 12,200 with neutrophilia.

On Gross examination, the ovary measured 4x3x2 cms. The external surface as well as the cut surface was haemorrhagic. On microscopic examination, plenty of chorionic villi lying dispersed in a background of haemorrhagic ovarian stroma were identified.

Case 3:

A 24 year old female. Grossly the ovary measured 3.5x2x1.5 cm was admitted on 7.04.14 . Usg was not be done in this patient, Her TLC -5,800/cu.mm & DLC were within normal limits. Detailed history could not be retrieved in this patient.

On Gross examination, the ovary measured 4x3.5x2 cms. The external surface and cut surface were haemorrhagic. Microscopy showed chorionic villi within a haemorrhagic ovarian stroma.

Discussion

Ectopic pregnancy can be classified as primary and secondary. Primary when ovum is fertilized while still within the follicle, secondary when fertilization takes place in the tube and the conceptus is later regurgitated to be implanted in the ovarian stroma. This can be either intrafollicular or extrafollicular. The intrafollicular type is invariably primary whereas the extrafollicular may be either primary or secondary in which case the ovarian tissue is absent in the gestational sac.

According to the Spiegelberg, the 6 criteria for a diagnosis of ovarian pregnancy are: (a) intact ipsilateral tube, clearly separate from the ovary; (b) gestational sac occupying the position of the ovary; (c) sac connected to the uterus by the ovarian ligament; and (d) histologically proven ovarian tissue located in the sac wall.

Ovarian pregnancy is more frequent with the use of intra-uterine device (IUD) [2] Assisted reproductive technology (ART) has become a risk factor for ovarian ectopic pregnancy (OEP). Risk factors such as pelvic inflammatory disease (PID) and prior pelvic surgery commonly seen in tubal pregnancies do not play a significant role in the etiology of ovarian pregnancies.

In two large series of OEPS, the risk factors implicated were found to differ. In a study of 110 OEPs by Po-Chun Ko; twenty-six (23.6%) patients used an IUD and 12 (10.9%) had undergone ART. OEP associated with ART increased in the last 10 years of the study period [3]. Francesco Plotti, et al reported a case of bilateral ovarian pregnancy in a young patient who had previously undergone intrauterine insemination (IUI) and controlled ovarian stimulation (COS) [4].

In another series of 49 cases where ovarian pregnancy was confirmed, the most common risk factors were found to be previous abdominal surgery for 19 cases and endometriosis for 16 cases. Four patients had a history of pelvic inflammatory disease, and only 2 patients had used an IUD. Huge uterine myoma was found in 2 cases, bicornuate uterus was found in 1 case, and arcuate uterus was found in 1 case [5].

Of the three patients reported by us, two did not have any known risk factor. Details were not available for the third patient and she was lost to follow up. According to the above-mentioned study the most common complaints are abdominal pain (42.9%) and vaginal bleeding (28.6%) [5]. The pain is generally nonspecific during early gestation and becomes worse as the pregnancy advances, sometimes culminating in cases of acute abdomen due to rupture of the gestational sac and placental disruption, resulting in haemoperitoneum. In the same study, interestingly, five asymptomatic patients were incidentally discovered to have an ovarian pregnancy during post-*in vitro* fertilisation (IVF) monitoring. The relation with ART was later supported by Ngu et al. [6], who reported that 28.5% of ovarian pregnancies were associated with IVF, indicating the high risk of ovarian pregnancy among women who undergo ART.

Although ovarian pregnancy is a rare event, awareness of this condition is important in reducing the associated morbidity and mortality.

An early diagnosis of ovarian pregnancy would help avoid serious complications and emergency surgery [7]. However, preoperative diagnosis is often difficult and the diagnosis is usually made during surgery [8]. Patient history, signs and symptoms, serum beta-hCG levels, and pelvic US examination may provide clues to the diagnosis.

In both our cases the ovarian pregnancy was clinically misdiagnosed as a tubal pregnancy. Though US was performed in one patient, a USG could not particularly distinguish the site of pregnancy. Patients with ectopic pregnancy often have low beta-hCG levels , and a higher incidence of tubal ruptures are reported in cases with decreasing beta-hCG [9]. This correlates with our second case who had a weakly positive pregnancy test, but exact levels were not tested.

Overall, ectopic pregnancy represents 1–2% of all pregnancies. 95% of these occur in the fallopian tube and only 1% are abdominal pregnancies. Only 0.5-3% of all ectopic pregnancies occur in the ovary. The overall incidence ranges from 1 in 2100 to 1 in 7000 pregnancies [10-12].

Of all cases of ectopic ovarian pregnancy, to the best of our knowledge, only 12 cases of advanced ovarian pregnancy have been reported so far based on a search carried out on PUBMED for the period from 1948 to 2015 [13-24].

The mechanism is thought to be fertilisation occurring outside the tube, followed by implantation within the ovary or the failure of follicular extrusion [24,25]. As ovarian pregnancy is very rare, the risk factors are not as well established. Possible risk factors are previous ectopic pregnancy, pelvic inflammatory disease, IUD use, endometriosis, previous abdominal surgery, uterine anomalies and assisted reproductive techniques (ART) [25–29].

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