CASE REPORT

Conception of an Ovarian Heterotopic Pregnancy

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ABSTRACT

Spontaneous heterotopic pregnancy is a rare clinical condition in which intrauterine and extrauterine pregnancies occur at the same time. The occurrence of an ovarian heterotopic pregnancy is a rare singular event comprising only 2.3% of all heterotopic pregnancies, extremely rare among naturally conceiving women. We present a case of a 30-year old patient with a spontaneously conceived heterotopic pregnancy. The patient was admitted to our hospital with left iliac fossa pain and amenorrhea. A transvaginal ultrasound scan showed an ovarian and an intrauterine heterotopic pregnancy. A laparotomy with left oophorectomy was performed at 10 weeks of gestation. The patient was followed up on regular antenatal visits. A healthy baby was delivered via spontaneous vaginal delivery at 38 weeks. Heterotopic pregnancy can occur in women of reproductive age in the absence of any predisposing risk factor; only with an early diagnosis and treatment the intrauterine pregnancy will reach viability with a greater chance of a favorable obstetric outcome.

KEY WORDS: *Spontaneous Conception, Adnexal Mass, Heterotopic Pregnancy*.

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INTRODUCTION

Heterotopic Pregnancy (HP) is defined as the simultaneous presence of intrauterine and ectopic pregnancy. First found during an autopsy in 1708[1], heterotopic pregnancy occurs in 1:30,000 of natural conceptions[1], while in assisted reproductive techniques (ART), the incidence is found to be as high as 1% [1] i.e. between 1:100 and 1:7000.[2,3]

In the last decades there has been a significant increase of ectopic pregnancy attributed to several factors, including higher incidence of pelvic inflammatory disease (PID) and the extended use of assisted reproductive technologies (ARTs) or any pelvic surgeries. Patients who are treated with ovulation-induction medication an incidence of 33/10,000 has been reported[4], whereas, it is extremely rare among women who conceive naturally. It carries a significant maternal morbidity and mortality due to the risk of ruptured ectopic pregnancy[5] hence, requires early diagnosis and management. Despite medical advances and techniques like ultrasonography[6], the rate of early diagnosis of heterotopic pregnancy hasn't much improved since 1970. Below we present a unique case history of heterotopic pregnancy. This case report stresses the importance of a holistic approach and thorough ultrasonography in managing these patients.

CASE

A 30 years old, Pakistani female (Para 2, Gravida 3) presented in the emergency department at our hospital on August 18, 2012, complaining of severe lower abdominal pain and feeling of being unwell. At this time, the patient gave history of 10 weeks of gestational amenorrhea and her last menstrual period was on June 9, 2012. She denied any vaginal bleeding, shoulder tip pain, urinary or bowel symptoms. She conceived spontaneously with no previous fertility treatments. She denied any history of sexually transmitted infections and had never used any contraception in the interim. She had no prior history of tubal or pelvic pathology, surgeries, ectopic pregnancies or miscarriages.

Figure 1: Transvaginal ultrasound of uterus showing an irregular intrauterine gestational sac of approximately 10 weeks

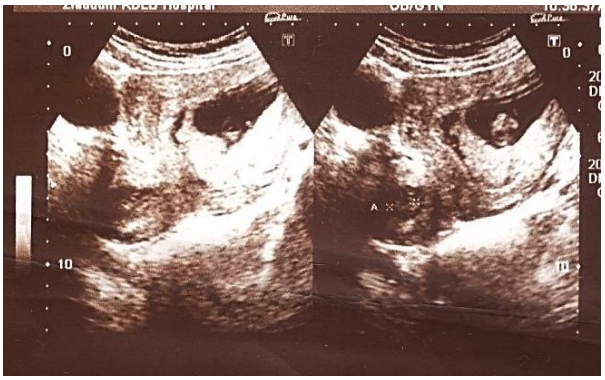


Figure 2: Transvaginal ultrasound showing left adnexal mass containing gestational sac with fetal pole

Her periods were regular and both prior pregnancies remained uneventful ending into spontaneous vaginal deliveries at term. Her past medical and surgical histories were unremarkable. She used no regular medications. She was not allergic to any drugs or food.

On examination, she was tachycardic at 110 beats per minute with a blood pressure reading of 122/62mmHg. Her abdomen was tense with severe suprapubic and left iliac fossa tenderness. A pelvic examination revealed a closed os and there was no bleeding from the cervix. On bimanual examination, the uterus was

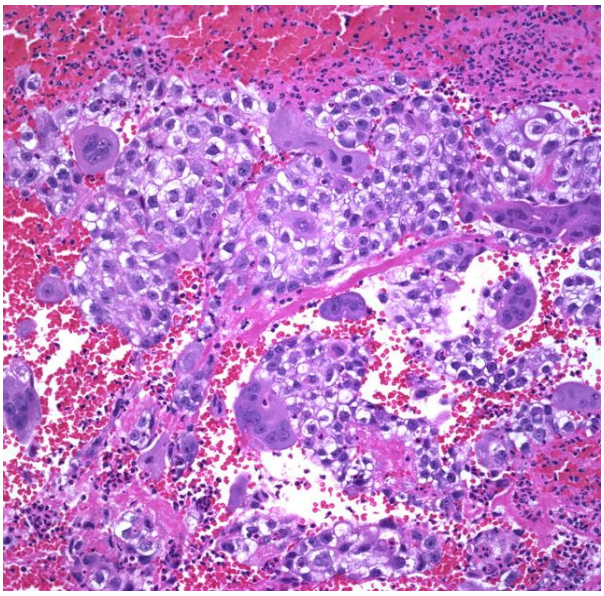
enlarged of about 10 weeks size with a tender left adnexa and positive cervical excitation tenderness. A palpable mass of about 3-4cm was felt in left annex. Her hemoglobin was 12.9g/dL, the haematocrit was 36.4% and white blood cell count was 4,900/mm^3.

Her serum β-hCG levels were 52,800mU/ml. A transvaginal ultrasound was done which revealed an intrauterine gestational sac according to 10 weeks of pregnancy, a yolk sac of 6mm and crown-rump-length (CRL) of 33mm with normal cardiac activity. The left adnexa showed another fetal pole with no cardiac flicker suggesting ectopic pregnancy. The right ovary along with fallopian tube was seen normal, measuring 3.0 x 2.1cm. No free fluid was seen in the pelvis.

The woman and her attendants were counseled and gave consent for an emergency surgical intervention. All preparations were done and the patient was shifted to the operation theatre a few hours later. The abdomen was opened via a Pfannenstiel incision. Operative findings revealed a normal bladder and enlarged uterus according to the gestational age. The right tube and ovary looked normal on gross appearance. The left tube was also normal but left ovary showed a cystic formation of about 2-3cm diameter. In the ovarian lower pole, a circular bleeding formation was seen, probably representing trophoblastic tissue. Left oophorectomy was done and ovarian pedicle was clamped, cut and ligated. Duration of surgery was 60 minutes and estimated intraoperative blood loss was 100ml. Prophylactic antibiotic was given and paracetamol was used for post-operative pain. The sample was sent for histopathologic examination and was confirmed as chorionic villi suggestive of an ovarian HP. The postoperative course remained uneventful and we discharged the patient on the 3rd post-operative day.

During the follow-up one week after the surgery an ultrasound scan was repeated and a viable intrauterine pregnancy was seen. She was prescribed progesterone support soon after surgery, along with the folic acid and calcium supplements. The patient continued her pregnancy and attended regular antenatal visits. She eventually delivered a healthy baby at term via spontaneous vaginal delivery.

Figure 3: High power view showing immature villous tissue consisting of cytotrophoblast and syncytiotrophoblast in an ectopic ovarian pregnancy



DISCUSSION

Health care professionals have been indoctrinated with the famous dictum by the French surgeon Henri Mondor (1885-1962) “Think Ectopic” when assessing the acute abdomen of a pregnant woman. However, when an intrauterine gestation is seen on ultrasonography, the dictum “Think Heterotopic” is often missed.

The co-existence of intrauterine and extrauterine pregnancy, also known as heterotopic pregnancy can occur in different forms: intrauterine pregnancy with tubal, abdominal, corneal, cervical or ovarian pregnancy. A previous review showed that most of the extrauterine pregnancies were located in the fallopian tube (72.5%).[6] An ovarian HP, as in our case, is a rare diagnosis with few reported cases. We reviewed the literature and only 3 cases of ovarian HP were spontaneous[7], while most of them were following clomiphene or ART use[11]. HP may be considered, in fact, a consequence of modern reproductive medicine. Whereas the frequency of spontaneous HP varies from 1:10,000 to 1:50,000, the wide spread use of ART may increase its incidence at nearly 1% in some series.

Spontaneous HP is a potentially fatal condition. Clinicians should maintain a high index of

suspicion in all patients presenting with amenorrhea, abdominal pain, adnexal mass, peritoneal irritation, and enlarged uterus, even if an intrauterine pregnancy has been confirmed. Suspicion should be higher in women with risk factors for an ectopic pregnancy and in low risk women who have free fluid in cul de sac with or without an adnexal mass with an intrauterine pregnancy.

An HP pregnancy is difficult to assess as pain and bleeding might be attributed to a threatened miscarriage. If ART is not involved, the index of suspicion of an HP is usually very low. This fact could cause delayed diagnosis, leading to serious complications. The presence of an intrauterine pregnancy, either viable or not, may actually mask the ectopic component of HP, resulting in delay of diagnosis. A simultaneous intrauterine pregnancy causes difficulties in the interpretation of ultrasound images as well. The ultrasonic visualization of heart activity in both intrauterine and extrauterine gestations is important for diagnosis but is rare.

Despite the advancement of TVS ultrasonography, identification of viable early intrauterine pregnancy before 5.5 weeks and ectopic pregnancy still remains challenging. Clinicians and sonographers tend to be lulled into a false sense of security when an intrauterine gestation sac is seen. This results in inadequate inspection of the adnexae and remaining structures during ultrasonography despite a strong initial clinical suspicion of ectopic pregnancy. Furthermore, during an ultrasound examination ovarian pregnancy is easily misdiagnosed as a corpus luteum. Therefore the early diagnosis of a HP is difficult. A review article in 2008[9] which analyzed 6 studies showed that transvaginal ultrasonography has a sensitivity of 74-84% in diagnosing ectopic pregnancy with specificity between 84-99.9%. However, ultrasonography is operator dependent. A comparative review of 192 cases of heterotopic pregnancies in 2007[6] showed that only 1/3rd of the cases were definitely diagnosed by ultrasonography pre-operatively like in this case study.

The only pathognomonic sign of HP is the simultaneous visualization of extrauterine and intrauterine fetal poles with cardiac motion. This occurs in only 10% of cases[10]. In our case, although the patient had no risk factors of ectopic pregnancy, previous pelvic inflammatory disease or clomiphene use, an accurate evaluation of clinical signs associated with a thorough ultrasound examination allowed a very early diagnosis.

B-hCG alone is not helpful in diagnosis of HP as subnormal hormone production by an ectopic pregnancy may be masked by the higher placental production from the intrauterine pregnancy. In our case, levels were 52,800mU/ml. Culdocentesis is an important aid in diagnosis when hemoperitoneum is present[11] as echogenic pelvic fluid is more important than anechoic fluid as it indicates the presence of peritoneal hemorrhage.

The standard treatment of ectopic pregnancy is surgery by laparoscopy or laparotomy depending upon the condition of the patient. Our patient was in severe pain so we had planned for emergency exploratory laparotomy. The ectopic component is removed during surgery, whereas the intrauterine component is expected to develop normally. The main aim of the surgery should be the preservation of the intrauterine pregnancy with minimal manipulation of uterus. The traditional method of treating an ovarian pregnancy is laparoscopic wedge resection or ipsilateral oophorectomy. Conservative or radical surgery may be done depending upon the condition of the contralateral tube. Laparotomy is preferred where laparoscopic facilities are not available as in our case. The gravid uterus should be handled carefully during the procedure. Fertility results have been found to be the same after laparoscopy or laparotomy. The preference of laparoscopic method over laparotomy is because of advantages of early post surgical recovery and minimal uterine handling and drying from exposure which may cause uterine irritability. The prognosis of intrauterine pregnancy is favorable in 60-70% of cases, irrespective of the surgical method used. An alternative treatment for HP is the use of methotrexate, but it is contraindicated in the presence of a viable intrauterine pregnancy as in our case.

HP pregnancy can occur in the absence of any pre-disposing risk factors, and the detection of an intrauterine pregnancy does not exclude the possibility of the simultaneous existence of an ectopic pregnancy. Therefore considering spontaneous pregnancies, every physician treating women of reproductive age should be

aware of the possibility of HP. A complete review of the whole pelvis including adnexa should be done at the time of ultrasound scanning. A high index of suspicion followed by an early surgical intervention can minimize maternal morbidity and preserve an intrauterine pregnancy

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