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CASE REPORT / OLGU SUNUMU

Diagnosis and clinical approach in primary ovarian ectopic pregnancy: A case report and review of the literature

Primer ovaryan ektopik gebelikte tanı ve klinik yaklaşımı: Bir olgu sunumu ve literatürün gözden geçirilmesi

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ABSTRACT

Primary ovarian pregnancy is a rare type of extrauterine pregnancy with an incidence of 0.5-1% of all ectopic pregnancies. The incidence ranges from 1/40000 to 1/7000 deliveries. We report a primary ovarian pregnancy in a spontaneus conception cycle in a 40-year-old woman with no predisposing factor. The patient was admitted to the emergency clinic with signs and symptoms of acute abdomen with heamoperitoneum , in a preshock state. An emergency laparotomy and wedge resection of the ovary was performed. Histopathologic examination showed chorion villi embedded in the ovarian tissue in multiple sections in different areas of the material. This is a discussion about this rare phenomenon, its ethiology, differential and clinical diagnosis and a review of literature.

Key words: Ectopic, pregnancy, ovary

ÖZET

Primer ovaryan ektopik gebelik oldukça nadir görülüp tüm dış gebeliklerin %0,5-1'ini oluşturur. İnsidansı 1/7000-40000 gebeliktir. Spontan bir siklusta hiçbir predispozan faktör olmaksızın 40 yaşında bir hastada oluşmuş bir primer ovaryan ektopik gebelik olgusu sunacağız. Hasta akut batın belirti ve bulgularıyla preşok durumda acil kliniğimize başvurdu. Transvajinal ultrasonografide 30 mm çapında kistik sol ovaryan kitle tespit edildi. Müracaatında hemoglobin seviyesi 8,4g/dl ve serum bHCG seviyesi 2548 mlU/ml idi. Acil laparotomi ve over kama rezeksiyonu uygulandı. Histopatolojik olarak alınan materyalin yapılan çoklu kesilerinde birçok alanda over dokusu içine invaze olmuş koryon villusları izlenmiştir. Bu nadir olgunun etyolojisi, ayırıcı tanısı, klinik tanısı hakkında bilgi verilerek literatür gözden geçirilmiştir.

Anahtar kelimeler: Ektopik, gebelik, over

INTRODUCTION

Primary ovarian pregnancy is one of the rarest types of ectopic pregnancy where the gestational sac is implanted within the ovary. It represents about 0.5-1% of all ectopic pregnancies and the incidence ranges from 1/40000 to 1/7000 deliveries. It is responsible for 10% of pregnancy related deaths. The diagnostic criteria were described in 1878 by Spiegelberg.[1] Unlike the tubal gestation ovarian pregnancy is associated with neither PID nor infertility. The only risk factor associated with the development of an ovarian pregnancy is the current use of IUD.[2,3] Misdiagnosis is common; because it is confused with a ruptured corpus luteum in 75% cases. Usually first clinical presentation is shock with hemoperitoneum requiring emergency surgery. In recent years, the incidence is increased because of the assisted reproductive techniques and wider use of IUD. We report a primary ovarian pregnancy in a spontaneous conception cycle in a 40 year old woman with no predisposing factor.

CASE REPORT

A 40-year-old, gravida 5 para 5 woman was admitted to the emergency service with a sudden onset of sharp low abdominal pain and faintness. She had no history of pelvic inflammatory disease, abortions, and use of intrauterine devices. She was pale. She had rebound tenderness on the left abdominal

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region. On bimanual vaginal examination the uterus was enlarged to 7-week pregnancy in size and a tender mass was palpated above the left fornix. On transvaginal ultrasonographic examination, the uterine cavity was empty, endometrium thickness was 13.1 mm and the right ovary was normal. In the left ovary, there was a ovarian cyst sized 25x30 mm with coagulum like sonographic appearance visualized around it and there seemed to be some amount of free fluid in the Douglas pouch. The blood pressure of the patient was 80/50 temperature was 37.0°C and pulse rate was 118/min being fillform in nature. In the complete blood count hemoglobin: 10.2 g/dL, hematocrit:33.1%, βhCG level was 2548 mlU/mL. With these clinical and laboratory findings the patient was prediagnosed as ectopic pregnancy and an emergency laparotomy was performed. On the exploration of the abdomen with a Pfannenstiel incision, the uterus was normal with no evidence of congenital anomaly, endometriosis or chronic inflammation. The right ovary and the right fallopian tube were also normal. The left fallopian tube with its fimbrial end was intact and normal; however, there was about 200 mL clotted blood in the abdominal cavity. The left ovary was bleedy and cystic in appearance and some amount of placental material was observed when the coagulum on the surface of the ovary was removed. There was no adhesion or pathology in the pelvic cavity. Wedge resection was performed to the left ovary. After the peritoneal cleaning, abdomen was closed. The postoperative hemoglobin was 8.4 g/dL, hematocrit was 27.2%.

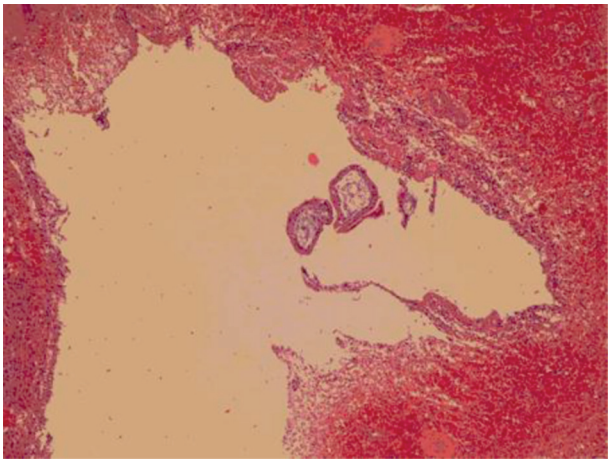

Figure 1. Chorion villi seen in the ulcerated kongested ovarian stroma H&E X40

The patient was discharged from the hospital on the third postoperative day without any complication. The βhCG level was 18.6 mIU/mL on the postoperative tenth day. Histopathological examination showed a primary ovarian ectopic pregnancy with chorion villi embedded in the ulcerated congested ovarian stroma (Figure 1).

DISCUSSION

Primary ovarian pregnancy is a rare phenomenon and its clinical diagnosis is very difficult. It deserves an additional afford not to be misdiagnosed as it leads to hemodynamic instability much more than the tubal pregnancies. Most cases represent as ovarian cysts.[4] Invasion of the trophoblastic cells of the ovarian tissue on the 6th day is followed by the invasion of the ovarian artery. Although ovarian pregnancies rupture by the 40th gestational day, reports of those progressing into the 3rd trimester even to live births have been established. Seki et al.[5] described an ovarian-pregnancy case of 30 weeks of gestation diagnosed at cesarean section. Shahabuddin and Chowdhury[6] reported a case of heterotopic pregnancy, the ectopic focus located in the ovary proceeding up to term. There was another case of an ovarian pregnancy by Belfar et al.[7] reporting a survived neonate until term. Studzinski et al.[8] reported an advanced ovarian pregnancy case with the retention of a dead fetus for more than a year. The risk factors of the tubal pregnancy do not correspond with the risk factors of the ovarian pregnancy. The only risk factor associated with the development of the ovarian pregnancy is the current IUD use. Raziel et al.[9] reported 20 series of ovarian pregnancy cases 18 of which had the history of IUD use. However in a series of 54 ovarian pregnancy study reported by Shiau et al.[10] only 13% of patients were current IUD users. In that study, they concluded that neither IUD use nor PID seemed to have a role in the occurrence of the reviewed ovarian pregnancies. Criteria for ovarian pregnancy diagnosis is described in 1878 by Spiegelberg (i) the fallopian tube on the effected side must be intact; (ii) the gestational sac must occupy the normal position of the ovary; (iii) the ovary must be connected to the uterus by the ovarian ligament; and (iv) ovarian tissue must be located in the sac wall.[1] All the criteria above are fulfilled in the case we presented. Ovarian pregnancies are classified as primary or secondary depending on

the implantation site of the conception material,[11] If the ovum is fertilized within the follicle it is called primary ovarian pregnancy; however, if the fertilization takes place in the fallopian tube and secondarily implants on the ovarian tissue it is accepted as secondary ovarian pregnancy. Primary ovarian pregnancy is considered to be due to an ovulatory dysfunction. William and Norris[12-13] stated that the tube must be intact and has no evidence of gestation and the ovarian tissue must be observed around the gestational sac at several positions to distinguish the primary from secondary ovarian pregnancy. The ovarian pregnancy is definitely diagnosed peroperatively. Misdiagnosis of this entity is common because it is confused with a ruptured corpus luteum in up to 75% of cases. It is debated that the frequency of ovarian pregnancy is underestimated since the medical therapy with methotrexate is used for suspected tubal pregnancies. This underestimation is balanced by more suspicion of the possibility of an ovarian pregnancy. Improvements in ultrasonography and use of more sensitive radioimmunoassay for βhCG detection lead to more accurate diagnosis of ovarian pregnancy cases. Although in the past culdocentesis was a very valuable tool for the diagnosis, hemoperitoneum can be visualized by vaginal ultrasonography. Cacciatore et al.[14] reported that with combined use of βhCG test and ultrasound the ectopic pregnancies can be diagnosed with 96% sensitivity and 100% specificity. Patients still present with circulatory collapse despite modern diagnostic modalities. The traditional method for management of an ovarian pregnancy is wedge resection or the removal of the effected adnexa by laparoscopy or laparotomy.[15] Systemic methotrexate admiration has become apparent in the recent years. But, it is not the first-line treatment of choice for ovarian pregnancy even if the patient is candidate meeting the criteria for medical treatment. In our case the patient was hemodinamically unstable so, an emergency laparotomy and wedge resection of the effected ovary was performed.

In conclusion the primary ovarian pregnancy is a rare entity. There are a few reports about a series of cases of ovarian pregnancies, the largest one including 54 cases. There might not be predisposing factors as in our case. The diagnosis is now easier because of the improved diagnostic modalities. Ultrasonographic appearance of an ovarian cyst in a patient with suspected ectopic pregnancy should imply us an ovarian pregnancy. The treatment is being more conservative due to earlier diagnosis. Despite these, many patients are admitted to the hospitals under hemodynamically unstable conditions when the conservative treatment is usually not possible. With the help of ultrasounds having better imaging quality it is now becoming more probable to make the diagnosis preoperatively.

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