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CASE REOPRT

TRIPLE HETEROTOPIC PREGNANCY; ECTOPIC TUBAL AND TWIN INTRAUTERINE PREGNANCY

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ABSTRACT

Introduction: Heterotopic pregnancy (HP) is defined as the coexistence of intra- and extra uterine gestations. We report a heterotopic triplet pregnancy with intrauterine twin gestations, presenting as hemoperitoneum, formerly misdiagnosed as corpus luteum.

Materials and Methods: A 33-year-old pregnant admitted to the emergency room (ER) with cramping pelvic pain. Physical examination revealed diffuse abdominal tenderness. Hemoglobin was 6.1 mg/dl. Ultrasound revealed twin IUP with positive fetal cardiac activities and hemoperitoneum ascending to liver.

Results: Laparoscopic investigation diagnosed the source of hemoperitoneum was left ampullar ruptured HP. Salpingectomy was performed. She was discharged after IUP viability was confirmed.
Unfortunately, 4 weeks later, she admitted to ER, diagnosed as missed abort. Dilatation and curettage was performed.

Discussion: IVF has been associated with an increased risk of both EP and HP. The incidence of HP is estimated to be 1 in 30,000 in spontaneous pregnancies. Early diagnosis of HP is often difficult because of the absence of clinical symptoms. However it is prudent to be suspicious in case of ART with pelvic pain, otherwise negligence lead to maternal mortality. In the literature case of triple heterotopic pregnancy comprising one tubal ectopic and twin IUP is really rare, our case comprises this rarity.

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INTRODUCTION

Heterotopic pregnancy (HP) is defined as the coexistence of intra- and extra uterine gestations. HP is a rare medical condition in obstetrics. The reported incidence varies, from 1:100 to 1:500 with the use of assisted reproductive technology (ART) to 1:30,000 pregnancies of natural conception (Berek, 2014). It is well known that ectopic pregnancy (EP) is the leading cause of pregnancy-related death in the first trimester (Main, 2015), and most patients with post-ART HP wish to remove the extra-uterine pregnancy while preserving the intra-uterine pregnancy (IUP). There has been a 300-fold increase in women conceiving with ART, including induction of ovulation, in vitro fertilization, and embryo transfer (Mihmanli *et al*., 2016). Therefore, the presence of an IUP can no longer be used to categorically exclude an EP in patients who have used ART (Mihmanli *et al*., 2016). We report a case of a ruptured tubal implantation of heterotopic triplet pregnancy with intrauterine twin gestations with positive fetal cardiac activities, presenting as acute abdomen with hemoperitoneum, formerly misdiagnosed as corpus luteum.

MATERIALS AND METHODS

A 33-year-old gravida 3 para 1 with C-section and living, abortus 1, pregnant woman admitted to the emergency room (ER) with a complaint of unbearable cramping pelvic pain and nausea. Two days before that admission to ER, she had admitted to the state hospital for her pain. She had been diagnosed as having twin pregnancy and corpus luteum, and she had been discharged. This time, she was consulted to our clinic, from ER, as having acute abdomen. Her blood pressure was 90/60 mmHg and pulse 108 beats/min. Physical examination revealed diffuse abdominal and cervical motion tenderness. Her hemoglobin and hematocrit were 6.1 mg/dl and 18%, respectively. The patient had history of IVF. She was 7 week-pregnant according to her last menstrual period. The patient had diffuse abdominal tenderness with left lower quadrant pelvic pain, and got worsen for 2 hours.

RESULTS

Vaginal ultrasound revealed 10-mm crown rump length, twin monochorial diamniotic IUP with positive fetal cardiac activities. Apart from this, US revealed diffuse hemoperitoneum below liver (Figure 1). Initially, emergent laparoscopy was planned with the pre-diagnosis of rupture of corpus luteum and heterotopic pregnancy. Laparoscopic investigation revealed, the source of hemoperitoneum was left ampullar ruptured heterotopic pregnancy, with the vision of oozing ruptured tuba uterina (Figure 2). Approximately 1200 cc hematoma was aspirated. Left salpingectomy was performed with laparoscopic sealing system (Ligasure ®, Covidien).

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She was given 2 unit of erythrocyte transfusion. She was discharged after two days of the operation uneventfully, her hemoglobin was 8.5 g/dL. IUP viability was confirmed. Unfortunately, 4 weeks later, she admitted to ER with the complaint of vaginal bleeding, diagnosed as twin intrauterin fetal demise. Dilatation and curettage was performed.

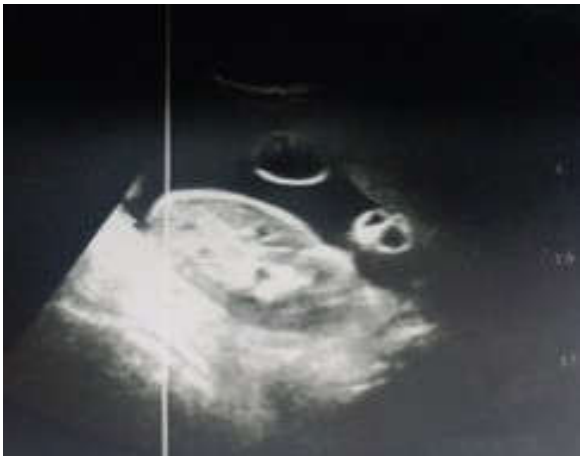

Figure 1. Ultrasound of hematoperitoneum

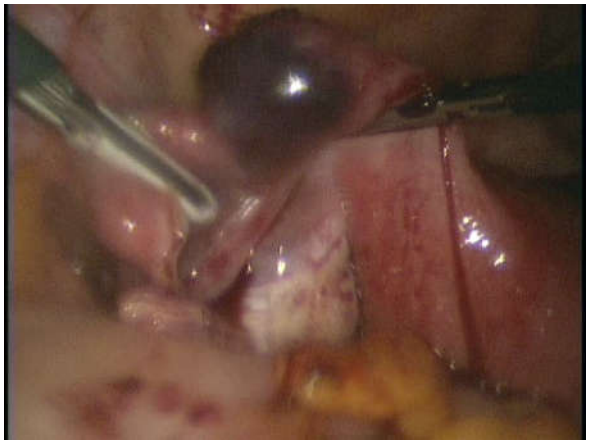

Figure 2. Laparoscopic view of left ampullar ectopic

DISCUSSION

IVF has been associated with an increased risk of both tubal EP and HP (Du *et al*., 2017). HP refers to the combination of an IUP and a concurrent pregnancy at an ectopic location. The estimated incidence of HP is dependent upon the rates of EP and dizygotic twinning. The incidence has been rising, mainly due to the increasing number of pregnancies derived from ART (Molinaro *et al*., 2018). The incidence of HP is estimated to be 1 in 30,000 in spontaneous pregnancies, but in pregnancies conceived using ARTs, it is reported to be as high as 1 in 100 (Schroeppel *et al*., 2006). In the literature case of triple heterotopic pregnancy comprising one tubal ectopic and twin IUP is really rare (Benítez, 2013; Sayin *et al*., 2003).

A HP should be considered in a patient with a viable IUP who is experiencing significant abdominal pain. Additional consideration should be taken when a patient has a history of ART, free fluid in the pelvis or adnexal mass on ultrasound, or has a rise in β-hCG after treatment (Molinaro *et al*., 2018). The presence of free fluid within the abdomen may be a sign of tubal rupture, but may be falsely labeled ascites associated with ovarian hyper stimulation syndrome (Rizk and Botros, 2010). The clinical presentation of HP is variable, 45% of patients being asymptomatic, 30% having abdominal pain and vaginal bleeding and about 25% presenting with abdominal pain but no vaginal bleeding (Rizk and Botros, 2010; Islam *et al*., 2017). Conservative treatment by locally injecting potassium chloride or hyperosmolar glucose is an option in HP (Felekis *et al*., 2014). Although Methotrexate can be used in corneal pregnancy (Sel *et al*., 2017), the use of methotrexate has detrimental effects on the residual IUP and is not an alternative in HP with intrauterine fetal cardiac activity (Cholkeri-Singh *et al*., 2007; Bugatto *et al*., 2010). The standard treatment for HP is surgery by laparoscopy or laparotomy, depending on the condition of the patient (Mihmanli *et al*., 2016). After diagnosis, the ectopic component is usually treated surgically. Most cases are treated by salpingostomy or salpingectomy by laparoscopy or laparotomy. In our case, we treated the patient by salpingectomy by laparoscopy. Early diagnosis of HP is often difficult because of the absence of clinical symptoms (Mihmanli *et al*., 2016). However it is prudent to be suspicious in case of ART with pelvic pain, otherwise negligence lead to maternal mortality.

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