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Spontaneous Heterotopic Pregnancy: A Case Report

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Abstract

Spontaneous heterotopic pregnancy is an uncommon condition that may result in maternal morbidity and mortality when not diagnosed early. In our case, a patient with severe pelvic pain and poor general condition presented to the emergency obstetric clinic and underwent transvaginal ultrasonography to find alive 7-week (crown-rump length: 6.5 mm) right-tubal pregnancy along with a live 7-week (crown-rump length: 8.2 mm) intrauterine pregnancy. Emergency laparotomy was performed due to the acute abdomen characterized by hemoperitoneum. And in term delivery was performed for the unharmed intrauterine pregnancy. Even if intrauterine pregnancy is detected in a patient presenting with abdominal pain, the possibility of heterotopic pregnancy should always be kept in mind and ruled out. Here we describe a case of spontaneous heterotopic pregnancy presenting with symptoms of acute abdomen in the light of the relevant literature.

Keywords: Heterotopic pregnancy; Ectopic pregnancy; Spontaneous pregnancy; Acute abdomen

Introduction

Heterotopic pregnancy (HP) is the presence of one or more ectopic pregnancies (EP)simultaneously with intrauterine pregnancy (IUP). Extrauterine pregnancy has a similar distribution of location a sectopic pregnancy. EP occurs most commonly in fallopian tubes but may also be ovarian, cervical, cornual, abdominal, or located in an old cesarean scar.

It was first reported by Duverney in an autopsy in 1708 [1,2]. In 1948, spontaneous HP was reported to be rare; the reported prevalence was 1/30000 in naturally conceived pregnancies. With the increased use of assisted reproductive techniques (ART) in recent years, however, prevalence was reported to reach up to 1/100 pregnancies conceived as a result of reproductive technologies in infertile women [3-10].

This condition, which should always be kept in mind in women of reproductive age, may cause abdominal pain, inguinal pain, vaginal bleeding, and acute abdomen in the first trimester; as well as serious maternal morbidity and mortality in cases of late diagnosis [4]. While late diagnosis has serious consequences, early diagnosis of HP is difficult. The most important diagnostic tool is transvaginal ultrasonography (TVS). About 70% of cases are diagnosed between the 5th and 8th weeks of the pregnancy, 20% between the 9th and 10th weeks, and only 10% after the 10th week [5].

Here we report a case of HP involving a 7-week live singleton IUP along with a ruptured, right tubal EP that occurred in a spontaneous cycle.

Case Report

A 31-year-old woman (gravida 1, para 0) presented to the emergency obstetric clinic at Okmeydanı Training and Research Hospital with pelvic pain and poor general condition, stating that she had no periods for the last 2 months and might be pregnant. The patient had no history of abdominal surgery, pelvic inflammatory disease, endometriosis, or trauma. Her blood pressure was 90/60 mmHg, pulse 100 per min, and respiration rate 24 per min. In her physical examination, she had widespread tenderness in both lower abdominal quadrants, guarding and rebound tenderness in the lower right quadrant. In the meantime, duplicate vascular access was established, and routine blood tests were ordered. In the vaginal examination, cervical movements were painful, and no bleeding was detected. In the transvaginal ultrasonography, along with a singleton IUP with positive fetal heartbeat and a crown-rump length (CRL: 8.2 mm) corresponding to 7 weeks, a right tubal EP with positive fetal heartbeat and a CRL (6.5 mm) corresponding to 6 weeks and 4 days and free abdominal fluid in Pouch of Douglas was detected (Figure 1). In addition to these findings, transabdominal ultrasonography (TAS) showed free abdominal fluid in the perisplenic and perihepatic areas (Figure 2). Doppler ultrasonography (DUS) revealed that both fetuses had heartbeats (Figure 3). Her hemoglobin level was 8.4 g/dl, hematocrit was 25%, leukocyte count was 17,000/mm^3, and her renal and liver function tests and coagulation parameters were within normal limits. An emergency laparotomy with diagnoses of

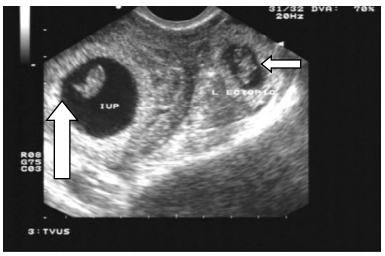

Figure 1: The TVS view of the IUP and right tubal EP. Small arrow: right tubal EPCRL: 6.5 mm; large arrow: IUPCRL: 8.2 mm.

Figure 2: The TAS view of the intrauterine and right tubal live fetuses.

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Figure 3: The DUS view of the intrauterine and right tubal fetuses and their heartbeats.

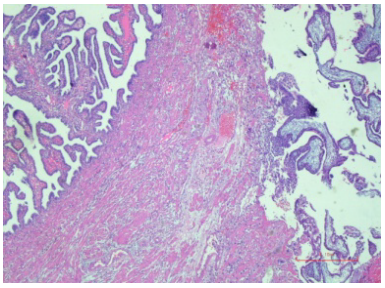

Figure 4: The natural appearance of the tubalepithelia and placental transition (H&E, 40 x).

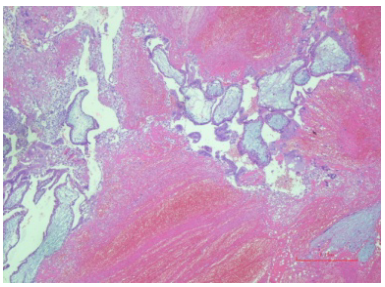

Figure 5: Villus structures surrounded by cytotrophoblast and syncytiotrophoblasts on the hemorrhagic, necrotic background (H&E, 40 x).

HP and acute abdomen secondary to hemoperitoneum was planned. Preoperatively depot progesterone was administered. Laparotomy was performed via a Pfannenstiel incision of the abdomen under general anesthesia. In exploration, approximately 1500 cm^3 of coagulated and defibrinated blood was observed in the abdomen. The uterus was enlarged. A ruptured EP compliant with 6 weeks of pregnancy was observed in the right tube, and the left adnexa was normal. Avoiding manipulation of the uterus, proper hemostasis was achieved by appropriate right salpingectomy. Following washing and aspiration using physiological saline solution, the abdominal layers were closed appropriately. After the operation, two-unit erythrocyte suspension was transfused along with one-unit fresh frozen plasma (FFP). Hydration was performed in the early postoperative period; the patient was started on oral micronized progesterone capsule (100 mg, 3 × 1) treatment. The patient was discharged on the fourth postoperative day without any complications. The TVS performed at that time showed the presence of fetal heartbeat in the IUP. Histopathological examination of the right salpinx removed during laparotomy confirmed the ruptured right tubal EP (Figures 4 and 5). The patient's routine pregnancy follow-ups were normal. A 3.490 gr live baby boy was delivered at the 39th week of the pregnancy with a cesarean section.

Discussion

Heterotopic pregnancy (HP) is rarely seen, difficult to diagnose, and can cause maternal morbidity and mortality. In HP, intrauterine pregnancy (IUP) is often accompanied by tubal ectopic pregnancy (EP). HPs involving EP in cesarean scars, bilateral tubal EP (triplet pregnancy), and complete molar pregnancy along with IUP have also been reported in the recent years with the frequent use of cesarean section and assisted reproductive techniques (ARTs) [6,7]. The other locations of accompanying extrauterine (ectopic) pregnancies are ovarian, cervical, cornual, and abdominal [9].

The predisposing risk factors for HP are similar to those of EP. These are primarily the history of tubal and other abdominal surgeries and also the history of pelvic inflammatory disease(PID), ART, intrauterine device (IUD) use, endometriosis, and Müllerian duct anomalies (MDAs). In the current case, however, none of these predisposing risk factors were present. HP is rarely seen in spontaneous cycles without any etiological factor. This is the second spontaneous HP reported in our clinic in the last 10 years.

Patient's complaints are variable. In their review of 66 HPs, Reece et al. listed the most obvious symptoms and findings for HP as abdominal pain, adnexal mass, peritoneal irritation, and enlarged uterus [1,11-13]. Vaginal bleeding is less common compared with EP. In the literature, the frequency of abdominal pain was reported as 55-83%, vaginal bleeding as 30%, and asymptomatic cases as 45% [11]. In one study, 70% of the HP cases were diagnosed in 5th to 8th weeks, 20% in 9th to 10th weeks, and 10% in the 11th week of pregnancy. Abdominal pain was observed in 83%; hypovolemic shock and acute abdomen findings were observed in 13% [3]. In the present case, the patient presented to the hospital with hemodynamic instability due to pelvic pain, widespread abdominal tenderness, and intraabdominal hemorrhage.

The b-hCG and progesterone levels can be used in diagnosis. Serial progesterone tests may determine the poor prognosis of pregnancy, but both are not suitable because of the coexisting IUP and these data do not have diagnostic value [6,13]. The best diagnostic tool for HP is ultrasonography, and transvaginal ultrasonography (TVS) has evident superiority over transabdominal ultrasonography (TAS) in this respect (93.3% and 50%, respectively) [12]. In addition, findings may be similar when IUP is associated with hemorrhagic corpus luteum cysts, including the ultrasonography view [8]. Emergency surgical pathologies such as acute appendicitis present with acute abdomen may also show similar findings as HP and should be considered in differential diagnosis [14-18]. In many of the cases, the EP is missed in presence of a simultaneous IUP and early diagnosis is not possible. Early diagnosis and appropriate treatment are important in terms of the morbidity and mortality of the patient and the fetus and in terms of the patient's future fertility demands. In a review covering the period between 1994 and 2004, 21 of 80 cases were diagnosed with preoperative ultrasonography and 59 with laparotomy or laparoscopy [3]. Although it had been reported in the previous series that only 10% of the cases were diagnosed pre-operatively, the sensitivity of ultrasonography was only 56%, and final diagnosis can only be made by surgery, it was reported in the later series that diagnosis with ultrasonography was 66% [14]. The recent increase in preoperative diagnosis is due to two factors: the increase in the resolution of the ultrasonography devices with the developing technology and the fact that HP is considered more often since ARTs are used more commonly. This report presents a case where spontaneous HP was diagnosed by visualization of tubal EP and IUP at the same time on preoperative TVS in a patient with acute abdomen.

The management of the treatment is mostly determined by the hemodynamic status of the patient, the localization of the EP, the expectation of the family regarding the IUP, and the experience of the surgeon. Emergency operation decision should be made in the presence of hemodynamic instability; this may be laparoscopy or laparotomy [15]. It was emphasized that care must be taken to protect the ovarian blood flow during operation, especially for the one with corpus luteum [19]. Although operative procedures might be considered in cases where the patient has stable hemodynamics, conservative methods such as local infusion of potassium chloride, hypertonic solution, or low-dose methotrexate might also be preferred [16]. Due to hemodynamic instability in this case, we performed laparotomy under general anesthesia. In the operation, right salpingectomy was performed, avoiding uterine manipulation in order to protect the IUP. Postoperative hydration and micronized progesterone treatment were performed to minimize the effects of manipulations and to reduce uterine contractions during surgery.

The prognosis of IUP depends on the choice of treatment modality. The IUP in HPs results in miscarriage 2 to 3-fold more likely than the IUP in normal pregnancies [17]. Recce et al. and Molloy et al. reported the ratio of pregnancies that continued and resulted in live delivery as 75% and 60%, respectively [19,20]. When laparotomy was performed for EP, the rate of abortion or stillbirth for IUP was reported as 9%, premature delivery as 16%, and in time delivery as 75% [14]. In addition, it is known that if the IUP manages to continue till the successful birth, the rate of poor outcomes such as low birth weight and preterm birth is not significantly different than those of normal IUPs, as is the case in this report [17].

Conclusion

The HP should not be overlooked in the first trimester in all pregnancies, especially in those presented with asymptomatic or abdominal pain and acute abdomen and in those conceived with the ARTs. During the evaluation of the IUP in the first trimester, adnexal areas should also be examined with TVS. Awareness, early diagnosis, and appropriate treatment approaches are important to prevent mortality and serious morbidity, to allow IUP to continue normally without complications, and to protect the patient's fertility.

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