

Case Report

Occult heterotopic pregnancy presenting with complex adnexal mass in a hemodynamically unstable 12 weeks' naturally pregnant woman

Gulseren Dinc¹, Suleyman Guven^{2,*}, Cavit Kart², Emine Seda Guvendag Guven²

¹ Medical Park Trabzon Hospital, Trabzon, Turkey.

² Karadeniz Technical University, School of Medicine, Department of Obstetrics and Gynecology, Trabzon, Turkey.

Abstract

Heterotopic pregnancy is very rare condition, it is estimated to occur in 1 in 30.000 natural pregnancies. Sometimes, the condition is a dilemma with respect to the diagnosis and the management. Biochemical tests (such as serial β -hCG level, serum progesterone measurements) and imaging studies may not be diagnostic in all time. Definitive diagnosis may only be reached via laparoscopy or laparotomy. A 33-year-old pregnant woman (gravida 1, para 0) was referred to our hospital with the complaint of severe abdominal pain and further evaluation of left adnexal complex cystic solid mass. Transvaginal ultrasonography and magnetic resonance imaging studies revealed 12 weeks' live intrauterine pregnancy and an additional left adnexal echogenic complex cystic and solid mass (10 cm x 6 cm in diameter) with high amount of free fluid in the peritoneal cavity. Emergency laparotomy confirmed the diagnosis of ruptured left tubal ectopic pregnancy with live intrauterine fetus. The clinicians should keep in mind the diagnosis of heterotopic pregnancy even in negative findings on clinical symptoms, laboratory evaluation, transvaginal ultrasonography and magnetic resonance imaging studies.

Key words:

Heterotopic pregnancy, complication, magnetic resonance imaging.

Introduction

Heterotopic pregnancy defines the presence of simultaneous pregnancies at least two different implantation sites. Most often these sites are a combination of intrauterine and ectopic (commonly tubal) pregnancies. Heterotopic pregnancy is very rare condition, it is estimated to occur in 1 in 30.000 natural pregnancies and 1 in 100 assisted reproductive technologies pregnancies (ART) [1, 2]. The increased incidence of pelvic inflammatory disease, the common use of ovarian stimulation agents and the advent of assisted reproductive techniques have contributed to the increasing incidence of heterotopic pregnancies in recent years [3]. Sometimes, the condition is a dilemma with respect to the diagnosis and the management. Biochemical tests (such

Article history:

Received 18 01 2015

Accepted 22 02 2015

* **Correspondence:** Suleyman Guven, MD
Karadeniz Teknik Üniversitesi Tıp Fakültesi,
Kadın Hastalıkları ve Doğum Anabilim Dalı, 61080, Trabzon, Türkiye.
Phone: +90 462 377 58 69,
E-mail: drsuleymanguven@yahoo.com

as serial β -hCG level, serum progesterone measurements) and imaging studies may not be diagnostic in all time. Definitive diagnosis may only be reached via laparoscopy or laparotomy. The surgical management may increase wastage risk of live intrauterine pregnancy. The aim of this case report is to report a case of heterotopic pregnancy presenting with complex adnexal mass in a hemodynamically instable 12 weeks' pregnant woman.

Case presentation

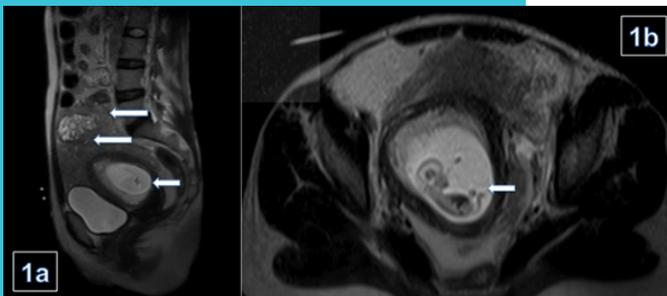
A 33-year-old pregnant woman (gravida 1, para 0) was referred to our hospital with the complaint of severe abdominal pain and further evaluation of left adnexal complex cystic solid mass. The pregnancy was unremarkable except for the complaint of small amount of vaginal bleeding. She had the history of spontaneous conception. History revealed that severe abdominal pain was started one day before admission and she admitted to local hospital, laboratory and sonographic evaluation revealed severe anemia (haemoglobine level: 7.2 g/dL) and com-

plex adnexal mass with high amount of free fluid in peritoneal cavity. Following two unit of blood transfusion the woman was referred to our tertiary center for further evaluation of severe anemia and complex adnexal mass. Initial evaluation of this patient revealed hypotension and tachycardia. Successive transvaginal and pelvic ultrasound examinations demonstrated a normally growing live fetus (12 weeks of gestation according to crown-rump length measurement) and an additional left adnexal echogenic complex cystic and solid mass (10 cm x 6 cm in diameter) with high amount of free fluid in the peritoneal cavity. Magnetic resonance imaging (MRI) studies also confirmed the sonographic findings (Figures 1a-b). Based on these findings and hemodynamically unstable condition, a preliminary diagnosis of torsion of ovarian mass and intraabdominal bleeding possible originating from left adnexal mass coexisting with live intrauterine pregnancy fetus was suggested. An emergency exploratory laparotomy revealed a hemoperitoneum of 2000 ml and a ruptured left tubal ectopic pregnancy. A left salpingectomy was performed. Two additional units of red blood cell suspensions were given during operation. The postoperative course was unremarkable and the woman was discharged on the fifth postoperative day. The pregnancy continued uneventfully and she delivered 3000 g live healthy infant at 39 weeks of pregnancy via vaginal route.

Discussion

Heterotopic pregnancy is a rare obstetric occurrence, especially in natural pregnancy. There is one fertilized ovum, which implants normally in the uterus, and one fertilized ovum, which implants abnormally, outside of the uterus. The advent use of infertility treatment agents and ART increased such rare obstetric entity. When the number of transferred embryo is increased, the risk of heterotopic pregnancy is also increased. However in natural conceptions, the incidence of heterotopic pregnancy is very low and still has been estimated to be 1 in 30,000 pregnancies [1]. The clinical diagnosis of this condition is also challenging. There are two possible ways for the occurrence of heterotopic pregnancy in natural cycles: (a) fertilization of two oocyte from a single coitus, (b) superimposition of an intrauterine pregnancy over an existing ectopic pregnancy (also known as superfetation) [4]. The appearance of cardiac activity and sonographic age may be discordant in heterotopic pregnancy. This condition may suggest that superfetation is indeed a mechanism in its development, with one pregnancy conceived earlier than the other [5]. In our case report, the discordant sonographic age of two different pregnancies since one of them was not diagnosed preoperatively with imaging studies, may also suggest the superfetation hypothesis. Clinically, the patients presented with non-specific symptoms, such as vaginal bleeding, abdominal pain and hemodynamic shock, or a complete lack of symptoms. Although combined serum β -hCG measurement and transvaginal ultrasound have significantly improved the diagnostic sensitivity of heterotopic pregnancy, location of a co-existing intrauterine pregnancy may lead to non-recognition of the heterotopic pregnancy in early pregnancy. In addition, heterotopic pregnancy is easily confused with other conditions, such as intrauterine pregnancy with hemorrhagic corpus luteum, adnexal mass and ovarian hyperstimulation syndrome after in vitro fertilization [6]. The uncertainty of clinical symptoms and laboratory/ imaging diagnostic modalities may limit the correct preoperative diagnosis. Sometimes, the correct diagnosis may be done via emergency exploration. In our case report, the clinical symptoms, laboratory evaluation, sonography and MRI were failed to elucidate the diagnosis of heterotopic pregnancy. The hemodynamic instability and large adnexal mass obligated us to explore this woman to reach the diagnosis of heterotopic pregnancy. The presence of a concurrent extrauterine pregnancy sometimes may be diagnosed by careful ultrasonography before

Figure 1.



T2 weighted image shows both intrauterine pregnancy (arrow) and complex cystic solid mass (arrows) located superior to uterus (1a). Axial T2 weighted image shows normal intrauterine gestational sac with fetus (arrow) (1b).

the onset of severe symptoms [7]. Transvaginal two-dimensional ultrasound, through observing a complex parauterine mass in association with a viable intrauterine pregnancy may not always give the opportunity to diagnose extrauterine pregnancy. However, under some conditions such as atypical ultrasonographic presentations, transvaginal sonography does not clarify the situation whereas MRI of the pelvis is able to do so. In this situation; MRI was of great importance in that it suggested that the mass had hematic content [8]. In our case the confirmation of hematic content of adnexal mass with MRI, clinical features, and unstable patient hemodynamic condition indicated emergency laparotomy. Heterotopic pregnancy can occur in the absence of any predisposing risk factors, and the detection of intra-uterine pregnancy does not exclude the possibility of the simultaneous existence of ectopic pregnancy [9]. Heterotopic pregnancies are usually diagnosed from 5 to 34 weeks of gestation. It was reported that 70% of heterotopic pregnancies were diagnosed between 5 and 8 weeks of gestation, 20% between 9 and 10 weeks and 10% after the 11th week [10, 11]. Our case also diagnosed at 12 weeks' of pregnancy. Its treatment is a challenge as serial beta-hCG is not useful in diagnosis or follow-up and medical management with methotrexate is contraindicated with an intrauterine pregnancy. Heterotopic pregnancies in hemodynam-

ically stable woman can be managed expectantly with strict monitoring and serial ultrasounds and the viable intra-uterine pregnancy can be saved. Laparoscopy or laparotomy with minimal manipulation of the uterus should be the standard form of treatment in these patients. Heterotopic pregnancy is possible with natural conception and the survival of the intrauterine fetus is feasible [1, 9]. In conclusion; the diagnosis and management of such emergency condition is real clinical dilemma. Even negative heterotopic pregnancy results for clinical evaluation, transvaginal ultrasound, the possibility of heterotopic pregnancy should remain within the differential diagnosis of any pregnant patient with either natural or assisted reproduction technology intrauterine pregnancy who presents with abdominal pain and/or clinical signs of ectopic pregnancy (such as hemodynamic instability).

Conflict of Interest

The authors declare that there is no conflict of interest regarding the publication of this paper

References

1. Barrenetxea G, L Barinaga-Reementería, A Lopez de Larruzea, JA Agirregoiakoa, M Mandiola, and K Carbonero. Heterotopic pregnancy: two cases and a comparative review. *Fertil Steril* 2007;87:417 e9-15.
2. Rojansky N and JG Schenker. Heterotopic pregnancy and assisted reproduction--an update. *J Assist Reprod Genet* 1996;13:594-601.
3. Chin HY, FP Chen, CJ Wang, LT Shui, YH Liu, and YK Soong. Heterotopic pregnancy after in vitro fertilization-embryo transfer. *Int J Gynaecol Obstet* 2004;86:411-6.
4. Wolf GC, L Kritzer, and C DeBold. Heterotopic pregnancy: midtrimester management. *Obstet Gynecol* 1979;54:756-8.
5. Hirsch E, L Cohen, and BR Hecht. Heterotopic pregnancy with discordant ultrasonic appearance of fetal cardiac activity. *Obstet Gynecol* 1992;79:824-5.
6. Yu Y, W Xu, Z Xie, Q Huang, and S Li. Management and outcome of 25 heterotopic pregnancies in Zhejiang, China. *Eur J Obstet Gynecol Reprod Biol* 2014;180:157-61.
7. Yamamoto R, H Murakoshi, Y Yamashita, Y Ejima, S Yoshida, and S Motoyama. Heterotopic pregnancy diagnosed before the onset of severe symptoms: case report. *Clin Exp Obstet Gynecol* 2013;40:445-7.
8. Sun SY, E Araujo Junior, J Elito Junior, LC Rolo, FF Campanharo, SG Sarmiento, et al. Diagnosis of heterotopic pregnancy using ultrasound and magnetic resonance imaging in the first trimester of pregnancy: a case report. *Case Rep Radiol* 2012;2012:317592.
9. Shetty SK and AK Shetty. A case of heterotopic pregnancy with tubal rupture. *J Clin Diagn Res* 2013;7:3000-1.
10. Tal J, S Haddad, N Gordon, and I Timor-Tritsch. Heterotopic pregnancy after ovulation induction and assisted reproductive technologies: a literature review from 1971 to 1993. *Fertil Steril* 1996;66:1-12.
11. Varras M, C Akkrivis, G Hadjopoulos, and N Antoniou. Heterotopic pregnancy in a natural conception cycle presenting with tubal rupture: a case report and review of the literature. *Eur J Obstet Gynecol Reprod Biol* 2003;106:79-82.