Heterotopic Pregnancy: A Rare Cause of Acute Abdomen. Report of a Delayed Case Diagnosis and Review of the Literature

Gazi **YILDIRIM**[1], Rukset **ATTAR**[1], Baki **EKCİ**[2], Cem **FIÇICIOĞLU**[1]

[1]*Hospital of Yeditepe University, Department of Obstetrics and Gynecology, İstanbul, Turkey*
[2]*Hospital of Yeditepe University, Department of General Surgery, İstanbul, Turkey*

Received 24 July 2008; received in revised form 11 August 2008; accepted 12 August 2008; published online 28 August 2008

Abstract

Heterotopic pregnancy is a potentially life threatining condition and an important cause of mortality and morbidity. Risk of heterotopic pregnancy is increased by assisted reproductive techniques (ART). We report such a case of heterotopic pregnancy after embryo transfer in a primary infertile 38-year old woman. Sixteen days after detection of intrauterine pregnancy by ultrasound, the patient was admitted to emergency service with severe abdominal pain. She underwent surgery and her preoperative diagnosis was undetermined intraperitoneal bleeding. Ruptured heterotopic pregnancy diagnosed after diagnostic laparotomy and salpingectomy performed at the same procedure. Her postoperative status and the course of pregnancy was unremarkable. A healthy baby was delivered at 38 week of gestation without any further complication. Heterotopic pregnancy is an important cause of acute abdomen after embryo transfer. Although it's diagnosis and management is challenging, with proper diagnosis and treatment, potential risks can be minimized and concomitant intrauterine pregnancy can be preserved.

Keywords: heterotopic pregnancy, acute abdomen, ectopic pregnancy

Özet

Heterotopik Gebelik: Akut Batının Nadir Bir Nedeni. Geç Tanı Konulmuş Bir Olgu Bildirimi ve Literatürün Gözden Geçirilmesi

Hayatı tehdit eden bir durum olan heterotopik gebelik mortalite ve morbiditenin önemli bir nedenidir. Yardımcı üreme tekniklerinden dolayı heterotopik gebelik riski artmıştır. Bu yazıda, 38 yaşında, birincil infertil bir kadında embriyo transferi sonrası oluşan bir heterotopik gebelik olgusu incelenmektedir. İntrauterin gebelik kesesi görüldükten 16 gün sonra acile ciddi karın ağrısı ile gelen olgu, nedeni bilinmeyen kanama sebebiyle laparotomi oldu. Laparotomi esnasında rüptüre ektopik gebelik saptanınca aynı seansta salpenjektomi uygulandı. İşlem sonrası dönemi ve gebelik süreci sorunsuz geçen hasta 38. haftada sağlıklı bir bebek doğurdu. Heterotopik gebelik embriyo transferi sonrası gelişen akut batının önemli bir nedenidir. Tanısı ve yönetimi karmaşık olsa da, uygun teşhis ve tedavi ile potansiyel riskler en aza düşürülür ve eşzamanlı intrauterin gebeliğin devamı sağlanabilir.

Anahtar sözcükler: heterotopik gebelik, akut batın, ektopik gebelik

Introduction

Heteroropic pregnancy is a potentially fatal disease. Diagnosis of these pathology is usually challenging. Incidence of heterotopic pregnancy has been reported as 1/8000-1/30 000 in natural conception (1). It may increase as high as 1% with assisted reproductive techniques (2).

Diagnosis of heterotopic pregnancy is a challenge not only for the obstetricians but also for other physicians who are following or treating the patient. We report a case of heterotopic pregnancy after embryo transfer. We discuss the emergency management of these cases and reviewed the literature.

Corresponding Author: Dr. Gazi Yıldırım
Barbaros Mah. Dereboyu Cad. Up Hill Court Sitesi B-b4 Daire: 109,
Kadıköy 34746 İstanbul, Türkiye
Phone : +90 216 578 42 00
GSM : +90 532 430 15 13
E-mail : gaziyildirim@gmail.com

Case Report

Mrs. FS, a 38-year old woman who has been married for fifteen years, presented in our clinic with prediagnosis of primary infertility. She had dysmenorrhea and occasional leukorrhea, other history was unremarkable. Her gynecological ultrasonography revealed a polycystic left ovary and more than ten antral follicles in her right ovary. Her husband had severe oligoastenoteratozoospermia. Her recent hysterosalpingography showed a sufficent intrauterine cavity and bilaterally normal tubal structures. Her BMI was 29 kg/m^2 and her basal hormone profile was normal. Other labarotory parameters and serological evaluation of the couple didn't reveal any other pathology. Her first antagonist cycle in our institution did not result in pregnancy then she received her second antagonist cyle three months later. Recombinant FSH 225 IU/day (Gonal F-Serono/Merk Laboratories) was started. When at least one leading follicle reached 14 mm in size, GnRH antagonist 0.25 mg daily injection (Cetrotide-Serono/Merk Laboratories) was added to the treatment and continued up till the hCG day. When leading follicles reached 18-20 mm diameter, 10 000 IU HCG (Pregnyl-Organon/Shering Plough) was administered and transvaginal ultrasound guided oocyte retrieval was performed 36 hours later. ICSI was performed to all oocytes. On day 3 after oocyte retrieval, 2 grade 2 and 1 grade 1 embryo were transferred. Luteal phase support was provided with daily progesterone vaginal gel (Crinone 8%-Serono/Merk Laboratories). Pregnancy test became positive in 12th day after transfer and pregnancy was confirmed with ultrasonography afterwards.

Four weeks after the transfer, patient had severe abdominal pain and presented at a local emergency service. On surgical examination her abdomen was noted to be rigid with a positive rebound. Also there was free fluid with coagulum in her abdomen. She was prediagnosed as intraabdominal hemorrhage of unknown etiology and operated with a sub- and supra-umbilical vertical incision. Ruptured tubal ectopic pregnancy was found in the surgery. A right salpingectomy was performed and 2 units of fresh erythrocyte suspension were given. She was discharged four days later. Her postoperative course was unremarkable. On her routine follow-up down syndrome risk was 1/75 on 11-14th week screening test and amniocenthesis was done for fetal karyotyping. Fetal karyotyping revelaed normal results. Diet-controlled gestational diabetes occurred during later her follow-up. Labor was started on 37-38th week spontaneously. Fetal distress occured during delivery anda 3200 g healthy baby boy was delivered by emergency cesarean section. There were no postoperative complications and patient was discharged without any problems except a reverse T incision.

Discussion

Today, assisted reproductive technologies are commonly used and sucessfully performed. This makes reproductive history an integral part of the approach in acute abdomen. In female patients presenting with acute abdomen, reproductive history should be carefully questioned and in cases of intrauterine pregnancy, an associated heterotopic pregancy should be kept in mind.

Heterotopic pregnancy is one of the most important risk of a successful *in vitro* fertilization. Besides the conventional risk factors, the procedures of assisted reproductive techniques (such as stimulation protocol, endometrial and ovarian reponse, embryo quality, transfer technique, number of transferred embryos and way of luteal support etc.) have been blamed. It is known that there is no ectopic pregnancy reported with transfers which are done after the third day (3). One idea advocates that the chosen embryo technique may increase the possibility of ectopic pregnancy in *in vitro* fertilization patients. Injection pressure during transfer, localization of transfer catheter tip and injection volume are thought to be possible initiators. Yovich et al. hypothysed that deep fundal transfer may increase ectopic pregnancy (4).

Management of heterotopic cases are difficult. Diagnosis and management are really challenging. Serial β-hCG measurements do not provide information because of the concomitant intrauterine pregnancy. Case series have shown that diagnoses have been made mostly after laparatomy (5,6).

All physicians who take care of women of reproductive ages should consider heterotopic pregnancy in the patients with acute abdomen. Laparoscopic removal of tubal or extratubal ectopic pregnancy does not have an unfavorable effect on the successful maintenance of the intrauterine pregnancy. With early diagnosis and intervention, intrauterine normal pregnancy's chance to continue increases to 70% (7).

Maintenance of intrauterine pregnancy is the suggested approach in the management of heterotopic pregnancy (8). Maternal hemorrhagic shock imperils the maintenance of intrauterine pregnancy. During surgery uterus should be preserved and any contact should be avoided. Laparoscopy is the primary treatment of choice in hemodynamically stabile patients (9). Most common surgery is salpingectomy by laparotomy. After this procedure abortus rate is 9% and early labor can occur in 16% of the patients. However, in 75% of the patients normal labor is expected (10). Because maintanence of pregnancy depends on the corpus luteum during these weeks, ovarian blood supply should be preserved during the surgery. Meticulous attention has to be paid to avoid the rupturing of corpus luteum during laparoscopy or laparatomy. In case of corpus luteum rupture, the progesterone supplementation should be started.

Bilaterel ectopic pregnancies due to assisted reproductive techniques have been reported (11). Therefore during surgery along with the abdomen, contralateral tubes should be carefully examined.

Laparoscopy should be primary choice in stable patients. There are a lot of studies reporting feasibility of laparoscopy during pregnancy (13). This technique allows early mobilization, therefore minimizes tromboembolic complications. However reliability of laparoscopy during pregnancy is not yet established. And the potential risks are: 1) Direct trauma to the uterus and fetus, 2) Impairment of uteroplasental perfusion due to increase in intraabdominal pressure, 3) Possible fetal acidosis from CO_2 absorption, 4) Carbonmonoxide intoxication risk to fetus as a result of laser or bipolar desiccation (13). In previously published studies, second trimester has been emphasized as the most appropriate period for laparoscopy.

In conclusion, acute abdomen requires a multidisciplinary approach. Especially in female patients presenting with acute abdomen, reproductive history should be carefully taken and if there is a pregnancy with assisted reproductive techniques, heterotopic pregnancy should be suspected. Laparoscopy should be the primary choice in stable patients. The decision of salpingectomy or salpingostomy should made during surgery and conditions including the damageing of the tube, the patients' hemodynamics and the experience of the theatre staff should be taken into account.

References

1. Ceci O, Caradonna F, Loizzi P et al. Ultrasound diagnosis of heterotopic pregnancy with viable fetuses. Eur J Obstet Gynecol Reprod Biol 1993;52:229-31.
2. Tal J, Haddad S, Gordon N, Timor-Tritsch I. Heterotopic pregnancy after ovulation induction and assisted reproductive technologies: a literature review from 1971 to 1993. Fertil Steril 1996;66:1-12.
3. Strandell A, Thorburn J, Hamberger L. Risk factors for ectopic pregnancy in assisted reproduction. Fertil Steril 1999;71:282-6.
4. Yovich JL, Turner S, Murphy A. Embryo transfer technique as a cause of ectopic pregnancies *in vitro* fertilization. Fertil Steril 1985;44:318-21.
5. Abdalla HI, Ahuja KK, Morris N, Lynn J. Combined intra-abdominal and intrauterine pregnancies after gamete intrafallopian transfer. Lancet 1986;2:1153-4.
6. Fisch B, Powsner E, Heller L et al. Heterotopic abdominal pregnancy following *in vitro* fertilization/embryo transfer presenting as massive lower gastrointestinal bleeding. Hum Reprod 1995;10:681-2.
7. Nguyen-Tran C, Toy EC. Case 3: obstetrical. Heterotopic pregnancy: viable twin intrauterine pregnancy with a viable right tubal ectopic pregnancy. J Ultrasound Med 2000;19:355-7.
8. Lemus JF. Ectopic pregnancy: an update. Curr Opin Obstet Gynecol 2000;12:369-75.
9. Berliner I, Mesbah M, Zalud I, Maulik D. Heterotopic triplet pregnancy: report of a case with successful twin intrauterine gestation. J Reprod Med 1998;43:237-9.
10. Reece ED, Petrie RH, Sirmons MF et al. Combined intrauterine and extrauterine gestation: a review. Am J Obstet Gynecol 1983;146:323-30.
11. Klipstein S, Oskowitz SP. Bilateral ectopic pregnancy after transfer of two embryos. Fertil Steril 2000;74:887-8.
12. Soriano D, Yefet Y, Seidman DS. Laparoscopy versus laparotomy in the management of adnexal masses during pregnancy Fertil Steril 1999;71:955-60.
13. Nezhat C, Seidman DS, Vreman HJ et al. The risk of carbon monoxide poisoning after prolonged laparoscopic surgery. Obstet Gynecol 1996;88:771-4.