

Contents lists available at ScienceDirect

Gynecology and Minimally Invasive Therapy

journal homepage: www.e-gmit.com



Case report

Heterotopic pregnancy in a natural conception cycle presenting as acute abdomen: A case report and literature review



S.H.M. Siraj*, W.W. Wee-Stekly, B.S.M. Chern

Department of Minimally Invasive Surgery, KK Women's and Children's Hospital, Singapore

ARTICLE INFO

Article history:
Received 5 November 2013
Received in revised form
9 February 2014
Accepted 21 February 2014
Available online 6 November 2014

Keyword: adnexal mass heterotopic pregnancy laparoscopy

ABSTRACT

Heterotopic pregnancy is the simultaneous development of an intrauterine pregnancy and an ectopic pregnancy. Although it is common with assisted reproductive technology (ART), this fatal condition rarely occurs in natural conception cycles, with a rate of just one in 30,000 pregnancies. A high index of suspicion can help in timely diagnosis and appropriate treatment. We report a case of heterotopic pregnancy in a 28-year-old woman presenting with signs and symptoms of acute abdomen at 8 weeks of amenorrhea. The diagnostic problems and literature were reviewed.

Copyright © 2014, The Asia-Pacific Association for Gynecologic Endoscopy and Minimally Invasive Therapy. Published by Elsevier Taiwan LLC. All rights reserved.

Introduction

Heterotopic pregnancy occurs when there are coexisting intrauterine and ectopic pregnancies. 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.¹ Although the extrauterine implantation site in heterotopic pregnancies is most commonly tubal (88.2%), abdominal implantation has also been observed (2.7%).² Defined as ectopic pregnancy that implants in the peritoneal cavity, abdominal implantation in heterotopic pregnancy poses unique diagnostic and therapeutic challenges with maternal mortality almost eight times greater than that of tubal pregnancies and 90 times greater than that of intrauterine pregnancies.³ It is a rare event with significant maternal morbidity and mortality. The incidence of heterotopic pregnancy is much higher in women who have undergone ovulation induction: it is no longer considered a rarity after treatment with ART.^{4,5} We present a rare case of heterotopic pregnancy with intrauterine gestation and with moderate hemoperitoneum in a natural conception.

Case report

A 28-year-old woman with 8 weeks of amenorrhea presented to the Emergency Unit with clinical features of acute abdomen. A

E-mail address: drshmsiraj@yahoo.com (S.H.M. Siraj).

urine pregnancy test was positive. She had no history of pelvic inflammatory disease, abortion, sexually transmitted diseases, intrauterine device use, abdominal surgery, or treatment for ovulation induction.

The patient was hypotensive at 80/60 mmHg. Pelvic examination showed an enlarged uterus with a closed cervix and tender left adnexum. Her blood type was O Positive. Her hemoglobin was 7.6 g/dL and her serum β human chorionic gonadotropin (hCG) level was 39518.6 international units (IU)/L. Formal transvaginal ultrasound of the pelvis by the radiologist demonstrated the presence of an intrauterine gestational sac (IUGS) within the endometrial cavity corresponding to 6 weeks and 2 days of gestation and a yolk sac with a fetal pole was seen within the IUGS. A few cystic structures were seen within the left ovary with echogenic material within them—these could represent left hemorrhagic ovarian cysts. Moderate to large amounts of hemoperitoneum were noted in the abdominal cavity.

In view of the acute abdomen, hypotension, and features of hemoperitoneum seen on ultrasound, a heterotopic pregnancy was suspected and diagnostic laparoscopy was offered. Emergency laparoscopy confirmed that the patient had an extrauterine ruptured left tubal pregnancy with 1.7 L of hemoperitoneum (Fig. 1). A left salpingectomy was performed (Figs. 2–6) and the intrauterine pregnancy was allowed to continue. Histology confirmed the tubal pregnancy. She was given one unit of blood transfusion. On the day of discharge, her hemoglobin was 8.7 g/dL and her β hCG was 46166.7 IU/L. Intrauterine pregnancy viability was confirmed.

Conflicts of interest: All authors declare no conflicts of interest.

^{*} Corresponding author. Department of Minimally Invasive Surgery, KK Women's and Children's Hospital, Singapore.



Fig. 1. Intrauterine pregnancy and adnexal mass with hemoperitoneum.



Fig. 2. Left adnexal mass mimicking corpus luteal cyst.



Fig. 3. Ruptured left fallopian tube.

Discussion

Simultaneous intrauterine and extrauterine pregnancy is called heterotopic pregnancy and it poses unique therapeutic challenges. It has been found in various forms but still is a rare event in natural conception cycles occurring in less than 1:30,000 pregnancies⁶;



Fig. 4. Products of conception.



Fig. 5. Removed left fallopian tube.



Fig. 6. After salpingectomy.

however, with the use of ART, the incidence rises to 1:100 and 1:500 pregnancies.⁷ Seventy percent of ectopic pregnancies are diagnosed between 5 weeks and 8 weeks of gestation, 20% between 9 weeks and 10 weeks of gestation, and 10% after 11 weeks of gestation.¹² Heterotopic pregnancy is often associated with major diagnostic difficulties in modern reproductive medicine. Reece et al⁸ regard the common presenting signs and symptoms for heterotopic pregnancy as abdominal pain, adnexal mass, peritoneal irritation, and an enlarged uterus. These presentations are, however, nonspecific and may be confused with other normal or abnormal pregnancy manifestations. Serial samples of β hCG measurements are often difficult to interpret because intrauterine pregnancy causes the β hCG concentration to increase appropriately.⁸ Early transvaginal sonographic examination is commonly performed in women with ART pregnancy. The identification of a heterotopic pregnancy, however, is still difficult because of the associated intrauterine pregnancy, which often leads to late detection of an extrauterine sac. 9 Abdominal paracentesis may diagnose a case of heterotopic pregnancy but even if it is negative a heterotopic pregnancy cannot still be excluded. Intrauterine gestation with hemorrhagic corpus luteum can simulate heterotopic gestation both clinically and on ultrasound 10 as in our case. Other surgical conditions of acute abdomen can also simulate heterotopic gestation clinically and hence contribute to the difficulty in clinical diagnosis.

A bicornuate uterus with gestation in both cavities may also simulate a heterotopic pregnancy. High-resolution transvaginal ultrasound with color Doppler is helpful because the trophoblastic tissue in the adnexa in a case of heterotopic pregnancy shows increased flow with a significantly reduced resistance index. 11 The diagnosis of heterotopic pregnancy, therefore, remains a diagnostic challenge.¹² There are a number of risk factors for heterotopic pregnancy, such as previous tubal damage, ectopic pregnancy, and ART techniques such as in vitro fertilization (IVF) and gamete intrafallopian transfer; pharmacological ovulation induction is also a reported risk. Heterotopic pregnancy can occur in the absence of any predisposing risk factors, and the detection of the intrauterine pregnancy does not exclude the possibility of the simultaneous existence of an ectopic pregnancy.¹³ Our case did not have any risk factors for heterotopic pregnancy; her ruptured tubal pregnancy presented as an acute abdomen. Heterotopic pregnancy is most likely to be missed in natural conception unless the ultrasound facility is available and the sonologist is aware and carefully screens the tubes; if the tubes are overlooked, rupture may present as an acute abdomen, which can progress to maternal shock, possibly leading to maternal mortality.

The goal of management of heterotopic pregnancy is to terminate the extrauterine pregnancy while taking precautions to minimize the possible threat to the intrauterine gestation. This can be done through laparoscopy or laparotomy with minimal manipulation of the uterus. Yet, the need to surgically remove the extrauterine pregnancy may pose harm to the intrauterine pregnancy. Jan et al¹⁴ reported three cases of heterotopic pregnancy with hemoperitoneum; in all of their cases, laparotomy was done with salpingectomy in two cases and salpingostomy in one case.

As a form of nonsurgical management of heterotopic pregnancy, selective potassium chloride has been applied successfully to cases of tubal, cervical, cesarean scar, and cornual or interstitial pregnancies with preservation of the coexistent intrauterine pregnancy. Luo et al 16 reported a success rate of 66.7% for preterm and term deliveries after treatment of heterotopic pregnancies. Currently, no data exist to enable prediction of expected risk of developmental complications involving neonates of heterotopic pregnancies. The intrauterine pregnancy associated with a heterotopic pregnancy is at increased risk for spontaneous abortion; in cases of ongoing pregnancy, it seems that the prognosis depends on the time of delivery.

This illustrated case did not have any risk factors for heterotopic gestation and presented with ruptured ectopic pregnancy with hemodynamic instability. A heterotopic pregnancy, although extremely rare, can still result from a natural conception. It requires a high index of suspicion for early and timely diagnosis. Prompt intervention can result in a successful outcome for the intrauterine fetus, ¹⁷ and it may also prevent tubal rupture and hemorrhagic shock, which can cause manal death.

References

- Berek JS, ed. Berek and Novak's Gynecology. 13th ed. Philadelphia, PA: Lippincott Williams & Wilkins; 2002:533–534.
- Rojansky N, Schenker JG. Heterotopic pregnancy and assisted reproduction an update. J Assist Reprod Genet. 1996;13:594–601.
- Atrash HK, Friede A, Hogue CJ. Abdominal pregnancy in the United States: frequency and maternal mortality. Obstet Gynecol. 1987;69:333–337.
- Dor J, Seidman DS, Levran D, Ben-Rafael Z, Ben-Shlomo I, Mashiach S. The incidence of combined intrauterine and extrauterine pregnancy after in vitro fertilization and embryo transfer. Fertil Steril. 1991;55:833

 –834.
- Marcus SF, MacNamee M, Brinsden P. Heterotopic pregnancies after in-vitro fertilization and embryo transfer. Hum Reprod. 1995;10:1232–1236.
- Ludwig M, Kaisi M, Bauer O, Diedrich K. Heterotopic pregnancy in a spontaneous cycle: do not forget it. Am J Obstet Gynecol Reprod Biol. 1999;87:91–103.
- 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. 1993;66:1–12.
- Reece EA, Petrie RH, Sirmans MF, Finster M, Todd WD. Combined intrauterine and extra uterine gestation: a review. Am J Obstet Gynecol. 1983;146:323–330.
- Soriano D, Vicus D, Schonman R, et al. Long-term outcome after laparoscopic treatment of heterotopic pregnancy: 19 cases. J Min Invas Gynecol. 2010;17: 321–324.
- Sohail S. Haemorrhagic corpus luteum mimicking heterotopic pregnancy. J Coll Physician Surg Pak. 2005;15:180–181.
- 11. Glassner MJ, Aron E, Eskin BA. Ovulation induction with clomiphene and the rise in heterotopic pregnancies: a report of two cases. *J Reprod Med.* 1990;35: 175–178
- 12. Shah Y, Zevallos H, Moody L. Combined intra and extrauterine pregnancy: a diagnostic challenge. *J Reprod Med.* 1980;25:290–292.
- Ljuca D, Hudić I, Hadzimehmedović A. Heterotopic pregnancy in natural conception – our initial experience: case report. Acta Clin Croat. 2011;50: 249–252
- **14.** Jan F, Naikoo GM, Rather MH, Sheikh TA, Rather YH. Ruptured heterotopic pregnancy: a rare cause for hemoperitoneum; report of three cases from Kashmir, India. *Indian J Surgery*. 2010;72:404–406.
- Yeh J, Aziz N, Chueh J. Nonsurgical management of heterotopic abdominal pregnancy. Obstet Gynecol. 2013;121:489–495.
- Luo X, Lim CED, Huang C, Wu J, Wong WSF, Chand NCL. Heterotopic pregnancy following in vitro fertilization and embryo transfer: 12 cases report. Arch Gynecol Obstet. 2009;280:325–329.
- Espinosa Picazo M, Alcántar Mendoza MA. Heterotopic pregnancy: report of a case and review of the literature. Ginecol Obstet Mex. 1997;65:482–486.