

Submitted
11.03.2018
Accepted
29.05.2018
Published
31.12.2018

Heterotopic pregnancy – how easily you can go wrong in diagnosing? A case study

Michał Ciebiera, Aneta Ślabuszevska-Jóźwiak, Kornelia Zaręba, Grzegorz Jakiel

1st Department of Obstetrics and Gynecology, Centre of Postgraduate Medical Education, Warsaw, Poland

Correspondence: Michał Ciebiera, 1st Department of Obstetrics and Gynecology, Centre of Postgraduate Medical Education, Czerniakowska 231, 00-416 Warsaw, Poland; tel. +48 607 155 177, e-mail: michal.ciebiera@gmail.com

DOI: 10.15557/JoU.2018.0052

Keywords

ectopic pregnancy,
heterotopic pregnancy,
twin pregnancy,
ultrasound

Abstract

Introduction: Heterotopic pregnancy is a rare, but potentially life-threatening pathology. The diagnosis of heterotopic pregnancy is still one of the biggest challenges in modern gynecology. The incidence of those pregnancies in natural conception is about 1:30000. **Case presentation:** We present an unusual case of a heterotopic pregnancy which was misdiagnosed in the first trimester as a dichorionic twin pregnancy. At 13 weeks of gestation, the patient presented with an acute abdomen, she was diagnosed with a heterotopic pregnancy, and therefore was operated on, with the excision of the ruptured fallopian tube and the ectopic pregnancy performed. **Discussion:** The presence of an intrauterine pregnancy does not rule out the presence of a coexisting ectopic pregnancy. Clinicians should always keep in mind that a heterotopic pregnancy may occur in a woman of reproductive age. Careful ultrasound scan of the uterus and appendages is a must in all women of reproductive age with clinical symptoms.

Introduction

Heterotopic pregnancy (HP) is defined as the presence of an intrauterine pregnancy (IUP) that coexists with an ectopic pregnancy (EP)⁽¹⁾. HP can occur in several forms, e.g. one-sided tubal pregnancy, bilateral pregnancy, etc. (all with IUP)^(2,3).

The diagnosis of HP is still one of the biggest challenges in modern gynecology. The incidence of those pregnancies in natural conception is about 1:30000⁽¹⁾, but in assisted reproduction it is much higher (1:100 – 1:500)⁽⁴⁾.

The most common risk factors for ectopic pregnancy include pelvic inflammatory disease, intrauterine devices, adhesions, a history of ectopic pregnancy, assisted reproduction techniques and ovarian hyper-

stimulation syndrome^(5,6). Also, for women covered by an assisted reproduction program there are additional factors, such as higher incidence of multiple ovulation, higher incidence of tubal malformation and/or tubal damage, and technical factors in embryo transfer which may increase the risk for ectopic and heterotopic pregnancy⁽⁷⁾. Our patient presented with a single, idiopathic adhesion, but the rest of her medical history was negative.

Most common clinical symptoms of HP include abdominal pain, an adnexal mass, peritoneal irritation and an enlarged uterus. In contrast to ectopic pregnancy, vaginal bleeding is an extremely rare condition⁽⁸⁾. HP can result in severe and potentially fatal complications, including intra-abdominal bleeding, uterine rupture, preterm delivery or miscarriage^(9,10).

Case presentation

A 34-year-old primipara was admitted to clinic at 13 weeks of gestation with severe abdominal pain. About 6 weeks earlier, the patient had been diagnosed with a dichorionic twin pregnancy (Fig. 1 A and Fig. 1 B). At 7 and 11 weeks of gestation, the patient had presented with abdominal pain, which was treated by drotaverine, and resolved permanently. The patient had been treated at a public hospital, where a transvaginal scan (TVS) had been performed, yet in the exam report data about both adnexa were unavailable. On admission to our hospital, the patient presented with acute abdomen symptoms. Clinical examination revealed painful, enlarged right appendages. Blood examination revealed anemia with a hemoglobin concentration of 9.1 g/dl. TVS revealed fluid in the pouch of Douglas, as well as two gestational sacs. The first gestational sac was an intrauterine pregnancy with a fetus of 70 mm in crown-rump length (CRL). The second gestational sac with a living fetus was located behind the posterior uterine wall, within the right fallopian tube. This fetus had a CRL of 64 mm, and presented with bradycardia. Due to severe symptoms and the suspicion of heterotopic pregnancy (HP), the clinical team decided to perform a diagnostic laparotomy. Hemoperitoneum was found and a right-sided heterotopic pregnancy was confirmed (Fig. 1 C). The right fallopian tube was attached to the lower part of the posterior uterine wall by a small adhesion. The adhesion was removed, and a salpingectomy was performed for ruptured tubal ectopic

pregnancy (Fig. 1 D). The patient was discharged after 6 days. Follow-up was performed at an outpatient clinic. The intrauterine pregnancy (IUP) proceeded without complications to a full-term vaginal delivery.

Discussion

According to Tal *et al.*, 70% of HPs are diagnosed between 5 and 8 weeks of gestation⁽¹¹⁾. The presence of an IUP complicates the diagnosis of a heterotopic pregnancy. Most clinicians think that the presence of IUP excludes an ectopic one, and after the diagnosis of IUP fail to examine the appendages at all. According to Talbot's data, 71% cases of HP had one risk factor and 10% had three or more risk factors. That is why a careful assessment of risk factors may lead to a correct diagnosis, but nothing can be done without a careful ultrasound scan⁽¹²⁾. TVS is a gold standard in diagnosis, yet it is effective in the hands of an experienced examiner. However, it has a low sensitivity – from 26.3% to 92.4%⁽¹²⁻¹⁴⁾. Difficulties can occur in differentiating HP from a corpus luteum cyst or hemorrhagic cyst⁽¹²⁾. Transvaginal sonographic examination is recommended in early pregnancy, especially in patients who conceived with the use of assisted reproduction techniques⁽¹⁵⁾. Lyu *et al.* advise to perform transvaginal scan in every woman after in vitro fertilization 4 weeks after embryo transfer⁽¹⁶⁾. The diagnostic role of human chorionic gonadotropin concentration in HP is debatable. In this case, the heterotopic pregnancy was mistaken for a healthy di-

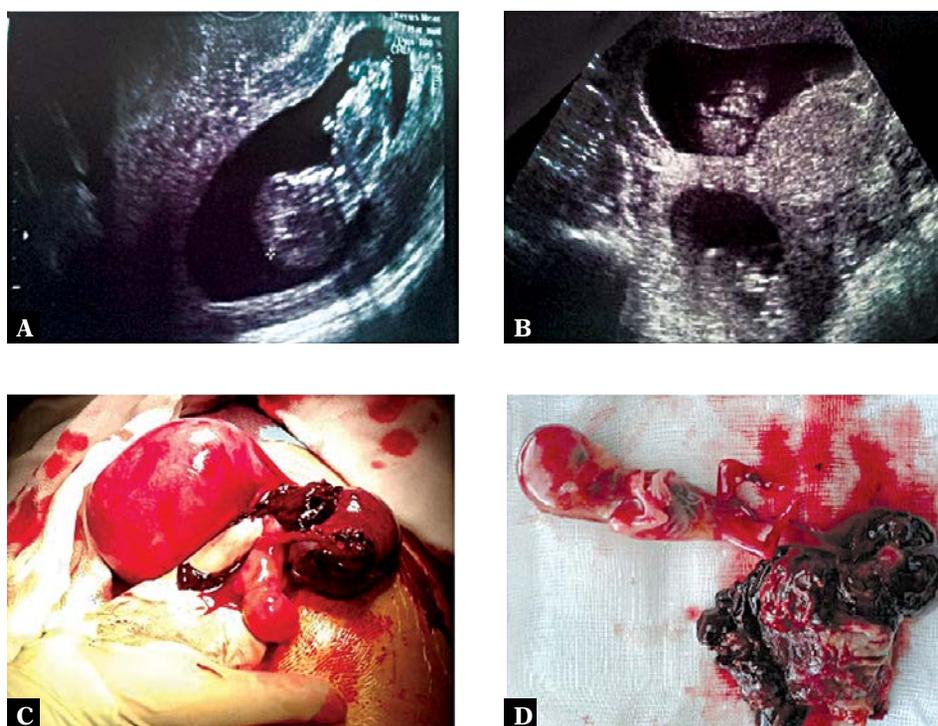


Fig. 1. Heterotopic pregnancy in ultrasound and during surgery. **A.** Scan at 11 weeks of gestation measuring the intrauterine fetus (different hospital). **B.** Scan at 11 weeks of gestation showing a “pseudo twin pregnancy” (different hospital). **C.** Ruptured right fallopian tube with the ectopic pregnancy during the surgery. **D.** Ruptured right fallopian tube with the ectopic pregnancy – after salpingectomy

chorionic pregnancy, probably because none of the sonographers examined the appendages.

The management of HP depends on the week of gestation. The key point of treatment is to preserve the IUP and resolve the EP⁽¹⁵⁾. In asymptomatic, stable patients expectant management might be considered^(13,17), but the risk of the rupture of HP is high. According to a study by Li JB, 20% of expectant management cases resulted in the rupture of HP⁽¹⁵⁾.

Surgery is still the most frequently chosen method of treatment. In most cases, it involves salpingectomy and depends on the actual clinical condition^(10,12). During surgery the manipulation of the uterus should be minimal, to preserve the IUP from complications. Data of 139 HP cases, treated mostly by surgery, revealed that the IUP survival rate was 66%⁽¹¹⁾. In women with unstable hemodynamic parameters, emergency surgery for HP rupture is strongly recommended⁽¹⁵⁾. Surgery involves mainly salpingectomy, salpingotomy or oophorectomy, but in some difficult cases it might also require even hysterectomy, with the risk of abortion in the group managed surgically being higher⁽¹⁸⁾. According to Li JB, the overall abortion rate in the group managed surgically was up to 14.8%⁽¹⁵⁾.

Ultrasonographically guided aspiration is a less invasive method with good effectiveness^(19,20). The problem is whether the location of the pregnancy is accessible by a needle. Potassium chloride or hyperosmolar glucose are agents of choice^(19,20). Pharmacological treatment with methotrexate should be avoided, due to the risk of its teratogenicity^(15,17), but there are some reports that show a good therapeutic effect of methotrexate⁽²¹⁾. Clinicians should be vigilant, as the incidence of this pathology rises, probably due to the

higher number of pregnancies after assisted reproduction⁽⁷⁾. Follow-up ultrasound scans are recommended due to the risk of failure or pregnancy rupture⁽¹⁵⁾.

The presence of an IUP does not mean that an ectopic pregnancy is absent. Clinicians should always keep in mind that a heterotopic pregnancy may occur in a woman of reproductive age. In the presented case, the pregnancy mimicked a dichorionic pregnancy and was thus unrecognized. This could have been avoided, if a detailed scan of the pelvic structures had been performed earlier. In our opinion, the appendages of each pregnant woman should be scanned, in whom clinical symptoms like abdominal pain, fluid in the pouch of Douglas or hypovolemic shock are present, or if the woman is in the high risk group. The surgical management of HP can result in a successful IUP and maternal outcome when early diagnosed.

Funding

This study was funded by the Centre of Postgraduate Medical Education, Grant Number 501-1-21-27-17.

Acknowledgements

This material is published with the patients' permission. This material has not been presented elsewhere.

Conflict of interest

All authors declare no conflict of interest.

References

- Hassani KI, Bouazzaoui AE, Khatouf M, Mazaz K: Heterotopic pregnancy: A diagnosis we should suspect more often. *J Emerg Trauma Shock* 2010; 3: 304.
- Wang PH, Chao HT, Tseng JY, Yang TS, Chang SP, Yuan CC *et al.*: Laparoscopic surgery for heterotopic pregnancies: A case report and a brief review. *Eur J Obstet Gynecol Reprod Biol* 1998; 80: 267–271.
- Fukuda T, Inoue H, Toyama Y, Ichida T, Uzawa Y, Monma M *et al.*: Bilateral tubal and intrauterine pregnancies diagnosed at laparoscopy. *J Obstet Gynaecol Res* 2014; 40: 2114–2117.
- Korkontzelos I, Antoniou N, Stefanos T, Kyparos I, Lykoudis S: Ruptured heterotopic pregnancy with successful obstetrical outcome: A case report and review of the literature. *Clin Exp Obstet Gynecol* 2005; 32: 203–206.
- Fatema N, Al Badi MM, Rahman M, Elawdy MM: Heterotopic pregnancy with natural conception; a rare event that is still being misdiagnosed: A case report. *Clin Case Rep* 2016; 4: 272–275.
- Jeon JH, Hwang YI, Shin IH, Park CW, Yang KM, Kim HO: The risk factors and pregnancy outcomes of 48 cases of heterotopic pregnancy from a single center. *J Korean Med Sci* 2016; 31: 1094–1099.
- Kirk E, Bottomley C, Bourne T: Diagnosing ectopic pregnancy and current concepts in the management of pregnancy of unknown location. *Hum Reprod Update* 2014; 20: 250–261.
- Reece EA, Petrie RH, Sirmans MF, Finster M, Todd WD: Combined intrauterine and extrauterine gestations: A review. *Am J Obstet Gynecol* 1983; 146: 323–330.
- OuYang Z, Yin Q, Xu Y, Ma Y, Zhang Q, Yu Y: Heterotopic cesarean scar pregnancy: diagnosis, treatment, and prognosis. *J Ultrasound Med* 2014; 33: 1533–1537.
- Yu Y, Xu W, Xie Z, Huang Q, Li S: Management and outcome of 25 heterotopic pregnancies in Zhejiang, China. *Eur J Obstet Gynecol Reprod Biol* 2014; 180: 157–161.
- 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.
- Talbot K, Simpson R, Price N, Jackson SR: Heterotopic pregnancy. *J Obstet Gynaecol* 2011; 31: 7–12.
- Li XH, Ouyang Y, Lu GX: Value of transvaginal sonography in diagnosing heterotopic pregnancy after in-vitro fertilization with embryo transfer. *Ultrasound Obstet Gynecol* 2013; 41: 563–569.
- Barrenetxea G, Barinaga-Rementería L, Lopez de Larrucea A, Agirregoikoa JA, Mandiola M, Carbonero K: Heterotopic pregnancy: Two cases and a comparative review. *Fertil Steril* 2007; 87: 417.e9–417.e15.
- Li JB, Kong LZ, Yang JB, Niu G, Fan L, Huang JZ *et al.*: Management of heterotopic pregnancy: Experience from 1 tertiary medical center. *Medicine (Baltimore)* 2016; 95: e2570.
- Lyu J, Ye H, Wang W, Lin Y, Sun W, Lei L *et al.*: Diagnosis and management of heterotopic pregnancy following embryo transfer: Clinical analysis of 55 cases from a single institution. *Arch Gynecol Obstet* 2017; 296: 85–92.

17. Baxi A, Kaushal M, Karmalkar H, Sahu P, Kadhi P, Daval B: Successful expectant management of tubal heterotopic pregnancy. *J Hum Reprod Sci* 2010; 3: 108–110.
18. Eom JM, Choi JS, Ko JH, Lee JH, Park SH, Hong JH *et al.*: Surgical and obstetric outcomes of laparoscopic management for women with heterotopic pregnancy. *J Obstet Gynaecol Res* 2013; 39: 1580–1586.
19. Lang PF, Weiss PA, Mayer HO, Haas JG, Hönigl W: Conservative treatment of ectopic pregnancy with local injection of hyperosmolar glucose solution or prostaglandin-F2 alpha: A prospective randomised study. *Lancet* 1990; 336: 78–81.
20. Goldstein JS, Ratts VS, Philpott T, Dahan MH: Risk of surgery after use of potassium chloride for treatment of tubal heterotopic pregnancy. *Obstet Gynecol* 2006; 107 (Pt 2): 506–508.
21. Sijanovic S, Vidosavljevic D, Sijanovic I: Methotrexate in local treatment of cervical heterotopic pregnancy with successful perinatal outcome: case report. *J Obstet Gynaecol Res* 2011; 37: 1241–1245.