

Contents lists available at ScienceDirect

Case Reports in Women's Health



journal homepage: www.elsevier.com/locate/crwh

Late presentation of a heterotopic pregnancy at 19 weeks of gestation leading to maternal collapse: A case report



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Keywords: Heterotopic pregnancy Ruptured heterotopic pregnancy Haemoperitoneum in preg	ARTICLE INFO	A B S T R A C T
· · · · · · · · · · · · · · · · · · ·	<i>Keywords:</i> Heterotopic pregnancy Ruptured heterotopic pregnancy Haemoperitoneum in pregnancy	Background: Heterotopic pregnancy is a rare form of pregnancy that involves implantation of simultaneous pregnancies at two sites. They are normally identified in early gestation when patients become symptomatic with vaginal bleeding or abdominal pain. Case Presentation: A 35-year-old woman (para 0, gravida 2) presented to the emergency department in hae-morrhagic shock at 19 weeks and 5 days of gestation. On assessment she was hemodynamically unstable and was found to have a large hemoperitoneum on ultrasound. She subsequently underwent an exploratory laparotomy and right salpingectomy for a suspected ectopic pregnancy. The postoperative period was uneventful, and the intrauterine pregnancy continued to term. Conclusion: Heterotopic pregnancy should be included in the differential diagnosis for women presenting with hemoperitoneum even beyond the first trimester.

1. Introduction

A heterotopic pregnancy (HP) is one that involves implantation of simultaneous pregnancies at two sites. Most frequently this involves a combination of an intrauterine pregnancy and a tubal ectopic pregnancy [1]. The estimated incidence of spontaneous HP is 1 in 30,000 pregnancies [2]. Whilst HPs are rare, increased use of assisted reproductive technologies (ART) has led to an increase in the overall incidence of HP [3]. Other risk factors for HP include pelvic inflammatory disease and previous fallopian tube pathology such as ectopic pregnancy.

HP can lead to serious complications and is potentially lifethreatening. Here we present a case of a spontaneous HP that presented with maternal collapse at 19 weeks of gestation. At surgery, a large hemoperitoneum was encountered and a right tubal mass noted. A right salpingectomy was performed and histopathology showed a tubal ectopic pregnancy. The patient had an uncomplicated recovery and proceeded to deliver a healthy baby girl vaginally at 39 weeks of gestation.

2. Case Presentation

A 35-year-old woman presented to the emergency department (ED) via ambulance following a witnessed syncopal episode at 19 weeks and

5 days of gestation, immediately following her fetal anatomy scan. She was para 0, gravida 2, having suffered one first-trimester miscarriage which was managed expectantly. She had no other medical history of note.

This pregnancy was conceived spontaneously. A dating scan at nine weeks of gestation showed a single intrauterine pregnancy with a normal fetal heart rate and a crown-rump length concordant with the last menstrual period. No adnexal masses were noted. A nuchal translucency scan performed at 12 weeks of gestation showed a single live intrauterine pregnancy with no adnexal masses noted. Aneuploidy screening indicated low risk and routine antenatal blood tests were unremarkable.

Her fetal anatomy scan showed an appropriately grown fetus with normal morphology. The placenta was posterior and 6.2 cm away from the internal os. No adnexal masses were noted and there was no pelvic free fluid seen.

On arrival at the ED, the patient was hemodynamically unstable with a pulse of 50 beats per minute (bpm), blood pressure (BP) of 40/ 20 mmHg, respiratory rate of 16 breaths per minute and oxygen saturation of 99%. She was maintaining her own airway and had a Glasgow Coma Scale (GCS) score of 15. Examination revealed a tender abdomen with a gravid uterus appropriately sized for dates. Speculum examination did not identify any vaginal bleeding. A focused assessment with

https://doi.org/10.1016/j.crwh.2021.e00331

Received 11 May 2021; Received in revised form 27 May 2021; Accepted 27 May 2021 Available online 31 May 2021

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sonography in trauma (FAST scan) was positive, identifying blood in the right upper quadrant, indicative of a 500–1000 mL hemoperitoneum. The fetal heart rate was bradycardic at 60 bpm.

Her blood tests on admission showed a pH of 7.38 and a lactate of 1.9 mmol/L and a haemoglobin level of 90 g/L with a normal coagulation profile (INR 1.1). A major transfusion protocol was activated and she received initial fluid resuscitation with two litres of crystalloid and two units of O-negative blood.

Given her haemodynamic instability and examination suggestive of major intrabdominal bleeding, the patient was taken for a category 1 exploratory laparoscopy with both obstetric and surgical teams present.

The patient had an arterial line placed in theatre and underwent a general anaesthetic. At laparoscopy a large hemoperitoneum (approximately 2000 mL) was noted. A washout was performed but no obvious source of bleeding was noted in the pelvis or upper abdomen (though the view of the posterior uterus and pouch of Douglas was obscured by the gravid uterus). A decision was therefore made to perform a midline laparotomy to further examine the patient for a source of bleeding. The abdomen was explored sequentially but no active bleeding point was noted to be distended, with a calcified mass measuring approximately 3×3 cm. A decision was made to perform a right salpingectomy as this was the presumed source of haemorrhage.

The patient received a further four units of packed red blood cells intra-operatively and was transferred to the intensive care unit (ICU) post-procedure. Fetal heart rate was normal at 150 bpm.

The patient was extubated on day 1 post-operatively with a haemoglobin level of 103 g/L and was transferred to the ward on the same day. Her recovery thereafter was uncomplicated, and she was discharged from hospital on day 4 post-operatively.

The specimen sent for histopathology consisted of a dilated right fallopian tube expanded with blood and chorionic villi, consistent with an ectopic pregnancy.

The patient was managed through a high-risk obstetric clinic for the remainder of her pregnancy. Given the concern for fetal hypoxia following a prolonged period of maternal hypovolaemia, a fetal MRI scan was performed at 28 weeks of gestation. This showed no evidence of any concerning neurological changes. Serial growth and wellbeing ultrasound scans throughout the pregnancy were also reassuring.

The patient subsequently presented to the delivery ward at 39 weeks of gestation with ruptured membranes in early labour. She proceeded to a spontaneous vaginal birth of a live female infant weighing 3165 g. Apgar scores were 9 at one minute of age and 9 at five minutes of age. Post-partum recovery was uncomplicated.

The mother reported the baby girl to be thriving at one year of age and meeting all her developmental milestones.

3. Discussion

This case report describes an unusual example of a late presentation of a heterotopic pregnancy, at 19 weeks of gestation. Over 90% of heterotopic pregnancies are diagnosed prior to 11 weeks of gestation, either as an incidental finding at first-trimester ultrasound, or due to pain or haemodynamic collapse secondary to intra-abdominal bleeding [4]. Initial presenting symptoms may be attributed to the intra-uterine pregnancy, thereby delaying diagnosis and putting the patient and the viable pregnancy at risk of poor outcomes.

This patient's dating scan at nine weeks of gestation and nuchal translucency scan at 12 weeks of gestation did not detect the presence of a right adnexal mass suggestive of a right tubal ectopic. A study by Li et al. which assessed the use of transvaginal ultrasonography (TVUS) to detect HPs in patients receiving an embryo transfer following in vitro fertilisation (IVF) showed TVUS to have a sensitivity of 92.4% and specificity of 100% for diagnosis. However, only 64.4% of HPs were diagnosed at the initial scan performed three to five weeks after embryo transfer. [4]

Whilst these rates suggest high levels of detection of HP with TVUS, these scans were performed in a tertiary centre, on patients undergoing IVF, who have higher rates of HP. A high index of suspicion is required to diagnose HP in a general setting, where rates are lower. HPs may frequently be missed on imaging due to both anchoring bias (whereby the clinician clings to the initial diagnostic impression and fails to adjust their impression in light of subsequent information) and satisfaction of search bias (whereby the clinician stops searching for abnormalities once a diagnosis perceived as being likely has been reached) [5].

Given her advanced gestation and normal first- and second-trimester ultrasound scans, a ruptured heterotopic pregnancy was not considered in the initial diagnosis on presentation. Consideration was given to more common presentations of acute hemoperitoneum in advanced pregnancy, including uterine rupture, hepatic and splenic rupture [6]. Whilst HP diagnosis at this late gestation is rare, there are several case reports in the literature [7,8]. It should therefore remain on the list of differential diagnoses in order to enable timely and appropriate management of these patients.

In this case, immediate recourse to surgery was required due the patient's haemodynamic instability and unclear diagnosis. A laparoscopic approach was initially taken due the minimally invasive nature of the procedure; however, it was difficult to clearly visualise the adnexa due to the size of the gravid uterus. Consideration should be given to the midline laparotomy approach as the primary mode of entry in the hemodynamically unstable pregnant patient, as this allows for adequate access to deal with multiple surgical pathologies [9].

For those patients with a HP who are hemodynamically stable, early detection and classification of HP in the first trimester may allow for conservative management, especially of those patients with an asymptomatic, small ectopic mass [10]. Larger ectopic masses or patients with pain or intra-abdominal bleeding will require immediate surgical management of the ectopic pregnancy. The ongoing live birth rate for the intrauterine pregnancy in both surgically and expectantly managed cases is around 70% [4,10]. The rates of both the extrauterine and the uterine pregnancies reaching term is lower. Reece et al. identified 13 cases of a total 589 reports [11].

This case shows the importance of maintaining a high index of suspicion for HP in the hemodynamically unstable pregnant patient beyond the first trimester of pregnancy. HPs may easily be missed on ultrasound in the general setting due to their rarity and the impact of anchoring and satisfaction of search bias at sonography. Provided there is appropriate resuscitation and definitive surgical management in a timely fashion, outcomes for mother and the viable intrauterine pregnancy are favourable.

Contributors

George Gabriel contributed to the design of the study, acquisition of information, and drafting of the manuscript.

Rebecca A. M. Taylor was involved in patient management and contributed to the design of the study, acquisition of information, and drafting of the manuscript.

Conflict of interest

The authors declare that they have no conflict of interest regarding the publication of this case report.

Funding

No specific grant from funding agencies in the public, commercial, or not-for-profit sectors supported the publication of this case report.

Patient consent

Written consent was obtained from the patient.

Provenance and peer review

This case report was peer reviewed.

References

- T.A. Molinaro, K.T. Barnhart, Cesarean scar pregnancy, abdominal pregnancy, and heterotopic pregnancy. UpToDate Web site. https://www.uptodate.com.acs.hcn. com.au/contents/abdominal-pregnancy-cesarean-scar-pregnancy-and-heterotopicpregnancy. Updated Apr 21, 2021.
- [2] E.A. Reece, R.H. Petrie, M.F. Sirmans, M. Finster, W.D. Todd, Combined intrauterine and extrauterine gestations: a review, Am. J. Obstet. Gynecol. 146 (3) (1983) 323–330, https://doi.org/10.1016/0002-9378(83)90755-x. 6344638.
- [3] J. Tal, S. Haddad, N. Gordon, I. Timor-Tritsch, Heterotopic pregnancy after ovulation induction and assisted reproductive technologies: a literature review from 1971 to 1993, Fertil. Steril. 66 (1) (1996 Jul) 1–12, https://doi.org/10.1016/ s0015-0282(16)58378-2. 8752602.
- [4] X.H. Li, Y. Ouyang, G.X. Lu, Value of transvaginal sonography in diagnosing heterotopic pregnancy after in-vitro fertilization with embryo transfer, Ultrasound Obstet. Gynecol. 41 (5) (2013) 563–569, https://doi.org/10.1002/uog.12341.
- [5] A. Sotiriadis, A.O. Odibo, Systematic error and cognitive bias in obstetric ultrasound, Ultrasound Obstet. Gynecol. 53 (2019) 431–435, https://doi.org/ 10.1002/uog.20232.

- [6] Y. Xu, Y. Zhou, J. Xie, X. Yin, X. Zhang, Intraperitoneal hemorrhage during pregnancy and parturition: case reports and literature review, Medicine (Baltimore) 98 (35) (2019), e16300, https://doi.org/10.1097/ MD.00000000016300.
- [7] W. Dendas, J.C. Schobbens, G. Mestdagh, L. Meylaerts, G. Verswijvel, C.
 V. Holsbeke, Management and outcome of heterotopic interstitial pregnancy: case report and review of literature, BMC Pregnancy Childbirth. 21 (2021) 61, https:// doi.org/10.1177/1742271X17710965.
- [8] D. Hassiakos, P. Bakas, G. Pistofidis, G. Creatsas, Heterotopic pregnancy at 16 weeks of gestation after in-vitro fertilization and embryo transfer, Arch. Gynecol. Obstet. 266 (3) (2002) 124–125, https://doi.org/10.1007/s004040100202. 121 97547.
- [9] A. Cohen, B. Almog, A. Satel, J.B. Lessing, Z. Tsafrir, I. Levin, Laparoscopy versus laparotomy in the management of ectopic pregnancy with massive hemoperitoneum, Int. J. Gynaecol. Obstet. 123 (2) (2013) 139–141, https://doi. org/10.1016/j.ijgo.2013.05.014.
- [10] C. Nguyen-Tran, E.C. Toy, Case 3: obstetrical. Heterotopic pregnancy: viable twin intrauterine pregnancy with a viable right tubal ectopic pregnancy, J Ultrasound Med 19 (5) (2000) 355.
- [11] E. Reece, P. Petrie, M. Sirmans, M. Finster, W. Todd, Combined intrauterine and extrauterine gestations: a review, Am. J. Obstet. Gynecol. 146 (3) (1983) 323–330.