CASE REPORT

Term delivery of a heterotopic pregnancy coexisting with ruptured tubal ectopic pregnancy: A case report

DOI: 10.29063/ajrh2022/v26i4.12

Kenneth Toby Maduako[1] and Vivian Onoh*

Consultant, Obstetrician Gynaecologist, University of Benin Teaching Hospital, Benin City, Nigeria[1]; Center of Excellence in Reproductive Health Innovation, University of Benin, Nigeria[2]

***For Correspondence:** Email: *kentobymaduako@gmail.com*

Abstract

Pregnancies that occur in two different implantation sites simultaneously is described as Heterotopic pregnancy (HP). In the current study, a case of term delivery of a heterotopic pregnancy coexisting with ruptured tubal ectopic pregnancy, diagnosed by ultrasound (US) and clinical examination findings, which was managed successfully. A 25 year old Nigerian female, gravida 4, para 2 (2 term gestation, 1 spontaneous abortion) presented at the Emergency room with acute abdominal pain associated with vomiting. She had 8 weeks amenorrhea and a positive pregnancy test three weeks prior to presentation. Transvaginal ultrasound scan revealed a six-weeks viable intrauterine gestation. A diagnosis of possible ectopic pregnancy was made. Further trans-abdominal ultrasonography imaging revealed viable intrauterine pregnancy with evidence of an echogenic mass measuring 6.5 x 7.5cm in the abdominal cavity with significant fluid collection, and both ovaries were visualized and separate from the mass. An emergency exploratory laparotomy with right salpingectomy was performed with minimal handling of the uterus and other pelvic structures. At 37 weeks and 5 days gestation, she had an elective C/S for a transverse lying fetus and delivered a live normal birth weight baby girl with a good Apgar score. *(Afr J Reprod Health 2022; 26[4]: 110-113).*

Keywords: Heterotopic pregnancy, term delivery, tubal ectopic

Résumé

Les grossesses qui se produisent simultanément dans deux sites d'implantation différents sont décrites comme une grossesse hétérotopique (HP). Dans la présente étude, un cas d'accouchement à terme d'une grossesse hétérotopique coexistant avec une rupture de grossesse extra-utérine des trompes, diagnostiqué par échographie (US) et résultats d'examen clinique, qui a été géré avec succès. Une femme nigériane de 25 ans, gravida 4, para 2 (gestation à 2 termes, 1 avortement spontané) s'est présentée aux urgences avec des douleurs abdominales aiguës associées à des vomissements. Elle avait 8 semaines d'aménorrhée et un test de grossesse positif trois semaines avant la présentation. L'échographie transvaginale a révélé une gestation intra-utérine viable de six semaines. Un diagnostic de possible grossesse extra-utérine a été posé. Une échographie trans-abdominale plus poussée a révélé une grossesse intra-utérine viable avec des preuves d'une masse échogène mesurant 6,5 x 7,5 cm dans la cavité abdominale avec une importante collection de liquide, et les deux ovaires ont été visualisés et séparés de la masse. Une laparotomie exploratoire d'urgence avec salpingectomie droite a été réalisée avec une manipulation minimale de l'utérus et des autres structures pelviennes. À 37 semaines et 5 jours de gestation, elle a eu un C/S électif pour un fœtus couché transversal et a donné naissance à une petite fille vivante de poids de naissance normal avec un bon score d'Apgar. *(Afr J Reprod Health 2022; 26[4]: 110-113).*

Mots-clés: Grossesse hétérotopique, accouchement à terme, extra-utérine tubaire

Introduction

Multiple-sited pregnancy, also referred to as heterotopic pregnancy, is a rare complication of pregnancy in which both extrauterine and intrauterine pregnancies co-exist. It is estimated at 0.6-2.5:10,000 pregnancies. Ovulation inductions, and assisted reproduction techniques (ART) such as in-vitro fertilization (IVF), Gamete intrafallopian transfer (GIFT) are known risk factors. Reports on heterotopic pregnancies have become more frequent in the scientific literature, since the advent of ART in the past decade including four recent case reports from Nigerian tertiary health institutes[1], each addressing combined intrauterine and extrauterine pregnancies.

We present a case of ruptured ectopic pregnancy with co-existing intrauterine pregnancy

where the ectopic pregnancy was treated surgically while the intrauterine pregnancy was carried to term and delivered vaginally. We also provide insights into the possible etiopathogenesis of this unusual presentation and review the literature.

Case report

A 25 year old Nigerian female, gravida 4, para 2 (2 term gestation, 1 spontaneous abortion) presented at the Accident and Emergency room of the Igbinedion University teaching Hospital, Okada, with acute abdominal pain associated with vomiting the morning of 16th June 2016. She described the pain as sporadic, present on all sides of the abdomen, and exacerbated by movement and body activity, and not relieved on rest. Vomiting was non-projectile and contained recently ingested meals. She had 8 weeks amenorrhea and a positive pregnancy test three weeks prior to presentation. Transvaginal ultrasound scan revealed a six-weeks viable intrauterine gestation. There was no history of infertility and pregnancy was spontaneously achieved.

On clinical examination, she was in severe painful distress and appeared ill looking;- temperature was 36.4^0c (97.52^0F);-blood pressure was 100/60 mmHg; heart rate, 92 beat per minutes; and respiration rate, 24 breath per minutes. Abdominal palpation revealed a full abdomen with marked tenderness confined to the suprapubic, iliac and lumbar region with rebound tenderness and guarding pelvic examination showed blood smeared vulva and scanty blood in vaginal vault The cervix was long and the cervical "os" closed. The uterus was slightly bulky, while no adnexal masses were palpable. A diagnosis of possible ectopic pregnancy was made. Packed cell volume was 24%;- microscopy of midstream urine yielded 5-10 WBCs per high power field with a bacterial count less than 10^5/ml; and urine culture yielded no growth at 48 hours.Further trans-abdominal ultrasonography imaging revealed viable intrauterine pregnancy with evidence of an echogenic mass measuring 6.5 x 7.5cm in the abdominal cavity with significant fluid collection, and both ovaries were visualized and separate from the mass.

The findings were suggestive of a ruptured right tubal ectopic gestation with a viable intrauterine pregnancy of about 8weeks gestation. On the strength of the clinical and ultrasound findings, an emergency exploratory laparotomy was performed with access through a sub umbilical midline incision.

Findings at surgery were as follows: Large haemo-peritoneum with blood clot, estimated at 1.2liter which contained a non-viable gestational sac with distorted tissues in the ruptured middle third of the right fallopian tube measuring about 8 x 6 cm with ragged edges, entire tube was edematous and congested looking dark brown. There was a bulky uterus measuring 12 x 10cm seen after evacuating blood at the ruptured end of the tube. The left tube was grossly normal in its entirety and both ovaries were polycystic and slightly enlarged. A right salpingectomy was performed with minimal handling of the uterus and other pelvic structures. Abdominopelvic cavity lavage was done gently with normal warm saline and hemostasis was adequately secured. The patient had a satisfactory recovery following surgery.

On the 5th day post-operatively, obstetric ultra-sonogram conducted showed a persistent viable Intrauterine fetus with normal heart tone. She remained stable and was discharged home on the 6th post-operative day. She was counseled to present immediately to hospital if any complaints, otherwise to come to the antenatal clinic in two weeks. She remained stable and continued on ANC as scheduled.

Histopathology of the specimen from the ruptured gestational sac and right fallopian tube showed organized blood clot containing both syncytial and Langerhans cells of chorionic villi and syncytial large giant cells, thus confirming diagnosis of Ectopic gestation in the tube. At 37 weeks and 5 days gestation, she had an elective C/S for a transverse lying fetus and delivered a live normal birth weight baby girl with a good Apgar score.

Discussion

Duverney first described Heterotopic pregnancy (HTP) in 1708[2,3]. Presently, the use of ART and fertility agents such as Clomiphene citrate can increase a patient's risk of heterotopic pregnancy. This is possibly due to the combined effects of presence of tubal disease, high level of estradiol and progesterone with hyperstimulation and the

subsequent simultaneous transfer of several embryos into the uterus with retrograde flow into the fallopian tubes[2,4]. Indeed, any factor predisposing a patient to an increased risk of ectopic pregnancy (EP) and/or multiple pregnancies can contribute to heterotopic pregnancy[2,3,5,6]. Spontaneous HTP has been reported in the scientific literature with an incidence of approximately 1 in 39,000. This is particularly so in certain areas now described as the 'TWIN BELT' with high rates of twinning and triplets. Most of the HTP cases are diagnosed late, resulting in significant morbidity and occasional mortality[2]. As no single investigation can predict the presence of a HTP, it should be suspected in any patient, presenting with lower abdominal pain in the early phase of an obvious IUP following fertility treatment or not, especially when they reside in the twin belt areas[3,7]. Often, abdominal and pelvic ultrasound scan fail to show an ectopic pregnancy or the ultrasound is misinterpreted because of the awareness of an existing intrauterine pregnancy (IUP)[2,6]. Demonstration of an IUP is not a reliable indicator for excluding an ectopic pregnancy[2].

The implication of increased multiple pregnancy rate are worrisome and includes early pregnancy losses, sometimes even prior to detection of pregnancy; late miscarriages, preterm birth and in our patient's case, heterotopic pregnancy with rupture tubal gestation and normal viable intrauterine gestation. All these justify the need to devote attention to improving diagnostic skills and techniques, especially in these areas, to help prevent or reduce the aforementioned complications. This review and case report aim to sensitize healthcare providers and researchers, particularly those within this defined 'twinning belt zone', of the need to focus attention on the appropriate diagnosis and prompt management of spontaneous HTP. Ectopic Pregnancy is normally visualized on Ultrasound Scan (USS) in three forms including "blob" sign, "bagel" (doughnut) sign or a gestational sac with a fetal pole[8]. But most USS reports make no comment of a search for coexistent ectopic pregnancy when assessing an intrauterine gestation, because a heterotopic pregnancy is still thought to be tremendously rare. For this reason, almost all ectopic pregnancies are diagnosed by excluding an intrauterine pregnancy[5]. The patient presented early in the pregnancy with a history of lower abdominal pain and vomiting, which are common symptoms of IUP. There was also a delay in the diagnosis of the EP component and as its presence was not identified until an EP rupture had occurred and the patient developed a significant hemo-peritoneum. Although the primary USS confirmed the presence of an IUP, it failed to identify the EP.

The management of HTP remains controversial. Surgical therapy has been the traditional mainstay but involves surgical and anesthetic risks to both the mother and IUP[6]. Treatment should be modified to the site of implantation and the least invasive treatment should be considered in order to preserve the concomitant pregnancy. Studies suggest that laparoscopic management is preferred over laparotomy in patients with a suspected EP, alongside a documented IUP because of minimal manipulation of the uterus. Therefore, laparoscopic salpingectomy is the standard approach and is the first line of treatment in the setting of a ruptured EP[3].

A non-surgical approach may be used safely and effectively to manage patients who are clinically stable and where a HTP is recognized relatively early in gestation. A successful non-surgical management of six cases of HTP using potassium chloride (KCl) injection into the tubal EP has been reported[6].

This case represents a spontaneous heterotopic pregnancy in a 22-year-old patient with no previous risk factors identified. It illustrates the importance of not immediately ruling out an IUP after identifying an ectopic pregnancy, and consistently placing a heterotopic pregnancy on the differential diagnosis. In the case of confirmed IUP and hemo-peritoneum, it is important to consider the possibility of ruptured heterotopic pregnancy, as was seen in the case presented here. In contrast, ectopic pregnancy undergoing surgical management, intrauterine devices such as uterine manipulator should be generally avoided due to the likelihood of coexistence of early intrauterine pregnancy that is not visualized by ultrasound. In the case of confirmed IUP with abdominal pain, further workup and close monitoring should be considered to rule out heterotopic pregnancy, especially after ART techniques. Fortunately, in this case, the IUP and extrauterine pregnancy were discovered simultaneously via ultrasound.

Conclusion

Heterotopic pregnancy remains a rare condition, however, appropriate investigations and prompt diagnosis should be done thoroughly in any pregnant woman presenting with alarming abdominal pain and adnexal abnormality. The patient should be thoroughly investigated using ultrasound and other appropriate imaging techniques available, to exclude this rare diagnosis and allow on-time proper management. More so, conservative surgical management allowed the viable intrauterine pregnancy to develop to term, ultimately leading to delivery. This case demonstrates that laparotomy can be a successful treatment modality for a heterotopic pregnancy, a finding supported by other authors[4].

References

1. Nabi U, Yousaf A, Ghaffar F, Sajid S and Ahmed MMH. Heterotopic Pregnancy - A Diagnostic Challenge. Six Case Reports and Literature Review. Cureus. 2019 Nov; 11(11): 6080.
2. Mistry BM, Balasubramaniam S, Silverman R, Sakabu SA and Troop BR. Heterotopic pregnancy presenting as an acute abdomen: A diagnostic masquerader. *Am Surg*,2000 66 (3): 307–308.
3. Perkins JD and Mitchell MR. Heterotopic pregnancy in a large inner-city hospital: a report of two cases. *J Natl Med Assoc*,2004 96: 363-366.
4. Hill J. Assisted reproduction and the multiple pregnancy: increasing the risks for heterotopic pregnancy. *J Diagn Med Sonogr*,2003 19: 258–260.
5. Mishra A, Youssefzadeh D and Parente JT. Heterotopic pregnancy. *Fem Pat*,1998 23: 39–42.
6. Scheiber MD and Cedars MI. Successful non-surgical management of a heterotopic abdominal pregnancy following embryo transfer with cryopreserved-thawed embryos. *Hum Reprod*,1999 14: 1375–1377
7. Archibong EI and Etuk SJ. Case report, Heterotopic pregnancy following induction of ovulation. *Trop J Obstet Gynaecol*,2002 19: 115–116.
8. Bignardi TT, Alhamdan D and Condous G. Is ultrasound the new gold standard for the diagnosis of ectopic pregnancy? *Semin Ultrasound CT MR*,2008 29 (2): 114–120.