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RESEARCH ARTICLE

ACUTE ABDOMEN AS A RESULT OF UNSUSPECTED SPONTANEOUS HETEROTROPHIC PREGNANCY WITH TUBAL RUPTURE: A CASE REPORT AND REVIEW OF THE LITERATURE

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ABSTRACT

Background: High level of suspicion of heterotrophic pregnancy (HP) is necessary, when women within their reproductive ages present with symptoms of acute abdomen. Although regarded as a rare phenomenon, increased incidence of heterotrophic pregnancy from spontaneous and ART (assisted reproductive technology) have been observed. It could lead to life-threatening condition, if diagnosis is delayed. Spontaneous heterotrophic pregnancy is the presence of two gestations simultaneously, with an estimated prevalence of 0.08% in normal conception. **Case Presentation:** A 33-year-old nulliparous university student, with previous history of 4 terminations of pregnancy, and a three-year history of secondary infertility from a broken marriage was presented to our accident and emergency department as a result of acute abdomen. Urgent urine pregnancy test with a careful ultrasound assessment led to the diagnosis of a HP. Emergency surgical intervention with supportive measures resulted in a successful feto-maternal outcome as the patient had a normal delivery of a male infant at term. **Conclusion:** We reported a case of heterotrophic pregnancy of a patient with issues of secondary infertility for a period close to three years, who presented with acute abdomen. Considering the presentation and the increase of this phenomenon from literatures, it is therefore imperative for an adequate advocacy about this possibility during pregnancy.

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INTRODUCTION

Historically, and presently incidences of Heterotrophic pregnancy (HP) are diagnosed accidentally, either when a patient presents with acute abdomen, or during suspected ruptured ectopic pregnancy. For instance, the first of its kind was described by Duverney in 1708 as an autopsy finding (Reece, 1983). Spontaneous heterotrophic pregnancy can be life threatening, even cause fetal wastage. Though regarded a rare type of pregnancy, currently it is on the increase; reasons like assisted reproductive techniques (ART), increase in some of the risk factors including ovulation inductions, frequency of Pelvic inflammatory diseases (PIDs), late pregnancies and in our environments' increased rate of unskilled termination of pregnancy, coupled with high rate of twinning has all resulted in an increased rate of ectopic pregnancy, which has inevitably increased the chances of HP (Rimpy, 2009; Tat *et al.*, 1993; D Adekanle *et al.*, 2009). HP is the presence of simultaneous gestations at two or more implantation sites (Ibrahim I Ayuba *et al.*, 2015). "Moreover, the presence of at least one risk factor for ectopic pregnancy in 71% of women with HP had been

reported" (Talbot, 2011), which was in line with this case report, as the patient had several terminations of pregnancy and history of secondary infertility. In the management of HP pregnancies, three modalities are available depending on the need of the patient and the circumstances involved, such as surgical (radical and conservative laparotomy or laparoscopic methods), medical and expectant management (Ibrahim, 2015 and Badejoko *et al.*, 2013). In the index case we opted for both the surgical and the expectant approach since the patient presented with an acute abdomen with a ruptured tubal ectopic pregnancy and a viable intrauterine pregnancy (Adekanle *et al.*, 2006; Badejoko, 2013 and Ikechukwu, 2013). The use of medical treatment modalities like, Hyperosmolar or potassium chloride injection and methotrexate were not considered, because the ectopic pregnancy component had ruptured with severe haemoperitonium, and the intrauterine component contained a viable foetus. Considering the need of the patient, evacuation of the intrauterine pregnancy was inappropriate (Adekanle *et al.*, 2006 and Ikechukwu *et al.*, 2013). Currently, the ongoing pregnancy is beyond the mid second trimester and it is stable, though the survival rate of the ongoing pregnancy is reported to be approximately 60-70% of the intrauterine pregnancy (IUP) (Adekanle *et al.*, 2006 and Badejoko *et al.*, 2013). However, the case of foetal loss after surgical

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intervention has been reported to be within the range of 40% as a consequence of the surgery (Ikechukwu, 2013). Some of the dangers of this phenomenon observed could be attributed to delays in diagnosis, most particularly in a resource deprived environment like ours, where patients' attitude to early antenatal care, and the use of early ultrasound scan is relatively poor (Odewale, 2008). The situation could be more catastrophic because of varying presentations and behaviors such as in asymptomatic patients or those with symptoms mimicking other intrabdominal acute conditions; like ovarian torsion, ruptured ovarian cyst, acute appendicitis etc. Therefore, high suspicion of HP and the use of ultrasound scan in very early state of any suspected pregnancy could contribute a lot in early diagnosis of HP; which could give room for better conservative management that will reduce cost, morbidity and mortality (Badejoko, 2013 and Li *et al.*, 2013). We report a case of HP in a natural conception cycle that presented with tubal rupture, with a viable IUP.

pressure of 120/60 mmHg, respiration rate 22cycles per minute. Abdominal examination revealed diffuse abdominal tenderness more at the right iliac and suprapubic regions with significant guarding and rigidity. Intra-abdominal organs could not be palpated due to tenderness, but bowel sounds were normal, pelvic examination was also differed due to pain. Patient was resuscitated with intravenous fluids and investigations were done; Urine for HCG (human chorionic gonadotropin) was positive, others included packed cell volume which was 26%. An urgent ultrasonography was done (Trans abdominal and transvaginal) which revealed a viable fetus CRL 23.37 = 9⁺³ weeks intrauterine gestation, with a similar right sided gestational sac with nonviable embryo, and another pelvic mass measuring 50x40 mm simple cyst (corpus luteum?). A moderate amount of fluid was present in the cul-de-sac and in Morrison's space. A diagnosis of heterotrophic pregnancy with tubal rupture was made. An informed consent was obtained from the patient for an emergency laparotomy.



Figure 1. Trans-vaginal ultrasound of uterus showing a regular intrauterine gestational sac of approximately 9⁺³⁻⁴ weeks, and a nonviable extra uterine pregnancy without fetal heart motion, haemoperitonium

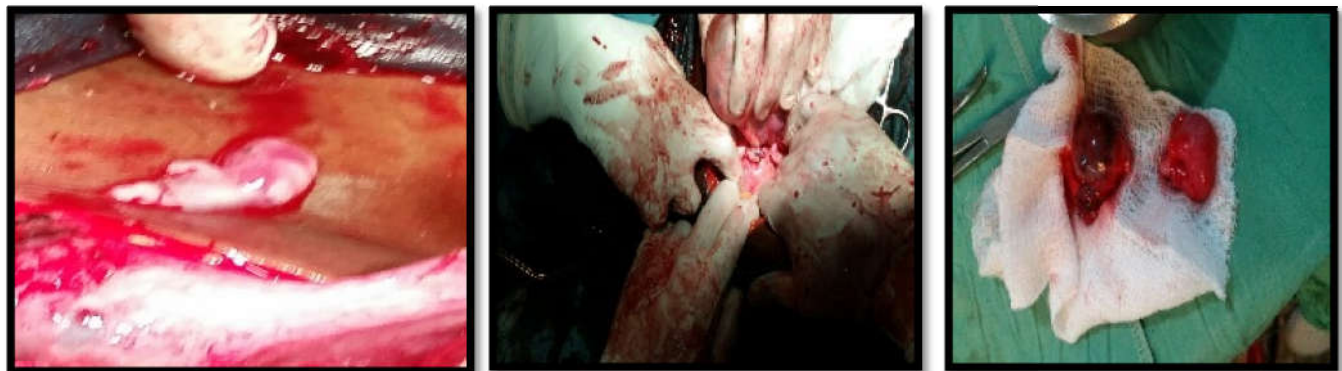


Figure 2. Showing intraoperative findings including nonviable abortum, corpus luteum, and part of ruptured interstitial ectopic part and isthmo-ampullary part of the right tube

Case Presentation

A 33-year-old final year university student, G5P0⁺⁴ were referred to our emergency department on account of acute abdomen from a private clinic after some intravenous and intramuscular medication without improvement of patient's condition. There was history of amenorrhea of uncertain period, as she could not remember her last menstrual period. There was also a three-year history of secondary infertility from a broken marriage. She previously had 4 terminations of pregnancy. There was no bleeding per vaginam, vaginal discharge, or urinary symptoms. On examination, she is a young woman who was in painful distress, a febrile, pale, dehydrated, with a pulse rate of 110 per minute and blood

The operational findings were a right ovarian cyst, a ruptured approximately 9-week right interstitial ectopic pregnancy, and about 10-week size gravid uterus, and the presence of approximately 1.1litres of hemoperitoneum. Right salpingectomy with removal of the hemoperitoneum and peritoneal lavage was performed. The surgery was done under general anesthesia and there was minimal handling of the uterus and the ovarian cyst to prevent uterine contractions and rupture. She was transfused with 2 units of blood and 4g Magnesium sulphate in 500ml/24hrs. Ringer lactate during and after the surgery and her postoperative period was uneventful. Other postoperative management includes; intravenous fluid for 24 hours, intramuscular pentazocine 30mg 6 hourly for 24 hours, intravenous ceftriaxone 1g 12 h-hourly for 48-hours,

and from the second day 200mg Duphaston (progestogen) till the end of the first trimester. The patient was discharged and followed-up regularly in the antenatal while on 12th of November 2016 at 13.29pm, she had spontaneous vaginal delivery of a live male infant at gestational age of 38weeks. Baby weighed 3.1 kilogrammes with Apgar's score of 9 in 1minutes and 10 in 10 minutes, length of baby 46cm. Mother and baby were discharged home 2nd day of delivery in good health.

DISCUSSION

Naturally occurrence of HP is still a rare gynaecologic emergency, and still represents high fetomaternal morbidity and sometimes mortality. Although in a natural conception cycle it is estimated to about 1 in 30,000 pregnancies, whereas the incidence of HP increases to as high as 1% with assisted reproductive techniques (Reece *et al.*, 1983 and Ikechukwu, 2013). This could be as a result of unexpected complications occurring during the transfer of embryos by ART techniques into affected tubes and peristaltic movements do not expel these embryos (Rimpy Tandon, 2009; Ibrahim, 2015; De Voe, 1948). Many authors have also reported an increase in ectopic pregnancy over the years due to increase of the risk factors which inevitably has influence on both conditions (EP and HP) (Russman, 2015). However, the actual incidence of HP in our environment is unknown and could be higher, due to higher risk factors for ectopic pregnancy in the environment such as: increased risk factors for PID increase in sexually transmitted diseases, high rate of unsafe termination of pregnancy; other reasons include increased birth rate, high incidence of twinning etc. (Adekanle, 2006). Some socio-cultural and traditional practices may also have contributed negatively in gathering proper data for scientific evaluation as the customs do not encourage autopsy and thorough medical investigation after sudden deaths of young people. Early diagnosis of HP is often difficult, even with the introduction of ultrasound because of the absence of some pregnancy related clinical symptoms, and none suspicion of the HP (Reece, 1983; Rimpy, 2009; Ibrahim, 2015 and Clayton, 2006). A recent literature review from 1994 to 2004 showed that out of 80 cases, 21 were diagnosed by ultrasound and 59 at laparoscopy or laparotomy (Reece, 1983 and Ikechukwu, 2013). This patient presented with only few non-specific features of ectopic pregnancy, like abdominal pain, two episodes of fainting attacks, and unknown period of amenorrhea, with history of secondary infertility. In spite of some of the patient's symptoms, any other abdominal catastrophe like (ruptured hemorrhagic corpus luteum, rupture spleen etc.) could have presented similarly, hence the initial management of the patient in the private hospital was in line with acute abdomen of unknown reasons (Russman, 2015 and Rimpy, 2009). Therefore, the presentation of this patient is not absolutely similar with the trend of presentation of ectopic pregnancy in Nigeria, which is usually seen as an acute emergency (Ibrahim *et al.*, 2015 and Odewale, 2008). Even though, Reece *et al.* "defined abdominal pain, adnexal mass, peritoneal irritation and an enlarged uterus as signs and symptoms suspicious of HP", only one of the features was clearly visible in the index case. HP is a rare condition and most patients present in the emergency department with symptoms of a rupture of ectopic component, which may reduce the preoperative diagnosis of HP if not thoroughly checked. The role of ultrasound examination most particularly the transvaginal ultrasound assessment of the whole pelvis in the index case was very vital as it provided the needed

information prior to the surgical intervention. As a result, conservative approach at surgery was necessitated; hence, the corpus luteum was handled with care to avoid any withdrawal effect on the IUP, because of the patient's need. The commonly adopted mode of management of the EP component of the HP is Laparotomy and laparoscopy, sometimes medical management (like the use of Methotrexate, hyperosmolar injection or direct potassium chloride injection) are also considered depending on the condition and sometimes with the desire of the patient (Aboyeji, 2001 and Varras, 2003). Though, this was a successful story as this pregnancy was carried to term and was concluded with a spontaneous vaginal delivery, however, it is estimated that, only approximately 60-70% success rates are possible in the IUPs. "Smith and Siddique reported 35% and 54% in 1971", and Tal *et al.* reported 66%, while Barrenetxea in another study reported 69% success rate where the IUPs progressed to term and were successfully delivered (Reece, 1983; Rimpy Tandon, 2009; Tat, 1993; Badejoko, 2013 and De Voe, 1948). Irrespective of the relative high success rate of HP it is still about 30% less likely to result in live birth than normal intrauterine pregnancy (Clayton *et al.*, 2006).

Conclusion

This pregnancy was a spontaneous conception from a new partner, there was a past history of abortions, secondary infertility, without history of previous pelvic inflammatory disease or abdominal surgery. Even though, incidence of heterotrophic pregnancy had been reported in the absence of any predisposing risk factors, the detection of an intrauterine pregnancy does not exclude the possibility of the simultaneous existence of an ectopic pregnancy, but in the index case there were two risk factors. Therefore, with the increase in the risk factors for ectopic pregnancy, high incidence of twinning, ovulation induction and ARTs in the country, irrespective of ultrasonography proof of the presence of an intrauterine pregnancy, high index of suspicion is required in all women within the reproductive age, presenting with any form of abdominal pain or severe abdominal discomfort. The situation is even more dangerous in this environment where, most of the pregnant patient's book late, and ultrasound scan done even much later. This present issue has confirmed the need for a high suspicion and advocacy of the possible of HP in all pregnancy until proved otherwise.

Consent: The patient gave a written consent for the case report to be published.

Abbreviations: HP: Heterotrophic pregnancy, PID: Pelvic inflammatory diseases, IUP: Intrauterine pregnancy, ART: Assisted reproductive technology, EP: Ectopic pregnancy.

Declarations

Acknowledgement: We do appreciate the contribution of the hospital nursing staffs, midwives, the laboratory staffs, all those who contributed to the successful completion of the case, and the management of the patient.

Competing interest: The authors declared that, none have competing interest concerning this case report.

Authors' contributions: EA, UC and AID treated the case and also contributed in writing the manuscript. II, OW and JI diagnosed the case ultrasonographically, physical and

laboratory examination of the case, secured consent and also involved in the general management of the case. Finally, all authors read and approved the final manuscript.

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