Spontaneous Heterotrophic Pregnancy with Tubal Rupture and Delivery of a Live Baby at Term: a Case Report

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Abstract

Background: *Heterotopic pregnancy is the coexistence of intrauterine and extrauterine gestations. It is associated with significant maternal morbidity and mortality particularly in low resource settings. Spontaneous heterotopic pregnancy is very rarely seen with documented incidence of 1 in 30,000 pregnancies. We present the first case of heterotopic pregnancy managed in our center with the delivery a live female baby at term.*

Methods/Result: *The case of a 25 year nulliparous lady who presented in our center with lower abdominal pains, amenorrhoea and an ultrasound report confirming an intrauterine pregnancy is presented. Examination revealed mild right iliac fossa tenderness, cervical motion tenderness and a bulky uterus. A trans-vaginal ultrasound scan confirmed a right fimbrial ectopic gestation. A right salpingectomy was performed. The patient subsequently had an uneventful antenatal period and spontaneous vaginal delivery of a live female baby at term. We also review literatures on heterotopic pregnancy and its management.*

Conclusion: *Spontaneous heterotopic pregnancy, a potentially fatal condition though rare can occur in our environment. Clinicians should maintain a high index of suspicion in all patients presenting with amenorrhoea and abdominal pains even if an intrauterine pregnancy has been confirmed and a thorough evaluation of the adnexae using a trans-vaginal ultrasound scan should be routinely performed in such cases.*

Key words: *Spontaneous heterotopic pregnancy, live baby, Uyo*

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Introduction

Heterotrophic pregnancy, a rare form of twin pregnancy is the coexistence of intrauterine and extrauterine gestations [1]. It is associated with significant maternal morbidity and mortality particularly in low resource settings where diagnosis is often difficult and might therefore be delayed. It was first reported in 1708 at an autopsy finding by Duverney in France. Spontaneous heterotrophic pregnancy is very rarely seen. The documented incidence rate is about 1 in 30,000 pregnancies [2]. Currently, with the increasing use of ovulation induction agents and assisted reproductive technologies (ART) the incidence of heterotopic pregnancy is reportedly increasing [1].

We hereby report the first case of heterotrophic pregnancy managed in our center which also resulted in the successful delivery of a live female infant at term. Our purpose is to remind practising clinicians about this very rare but potentially fatal gynaecological emergency and review literature of its management.

Case Report

Miss UTI a 25 year old nulliparous lady presented at the gynaecological clinic of the University of Uyo Teaching Hospital (UUTH) Uyo on the 3rd of October 2008 with complains of abdominal pains of 12 days duration and amenorrhoea of 8 weeks duration. Her last menstrual period (LMP) was on the 3rd of August 2008. There was no history of vaginal bleeding, contraceptive use, surgery, induced or spontaneous abortions. She had not been treated for any pelvic infection in the past. She was a University student and was single. She had first presented in a private clinic in Lagos where she was admitted for 2 days and was given some injections and intravenous infusions. A pregnancy test was also done which was positive and an ultrasound scan (USS) showed an intrauterine gestation. She was then referred to a local general hospital but preferred to present at UUTH because her relations resided in Uyo.

On examination, she was not pale, was afebrile and anicteric. The blood pressure and pulse rate were normal. There was mild tenderness in the right iliac fossa. The uterus was bulky, with mild cervical motion tenderness and right adnexal tenderness.

A repeat trans-abdominal USS showed a bulky uterus with a gestational sac and a live fetal node, a crown-rump length of 10 millimeters (mm) and a gestational age of 7 weeks and 1 day. There was a right adnexal

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mass, with an anechoic area within it and an indistinct outline. There was also free fluid collection in the hepatorenal fossa, paracolic gutters, and anterior and the posterior cul-de sac. Intestinal loops were seen to float within it. (Figure I). A transvaginal USS then showed a large well circumscribed gestational sac within the right fallopian tube with a fetal node within it. A diagnosis of right ectopic pregnancy coexisting with an intrauterine gestation was made.

The packed cell volume (PCV) was 32% while urinalysis revealed no abnormality. An exploratory laparotomy was offered and the patient consented. At surgery, the abdomen was entered through a Pfannenstiel incision. There was haemoperitonuem of approximately 500ml and the uterus was found to be enlarged to about 10 weeks gestational size with a right fimbrial ectopic gestation bleeding from its margins (figure II). The left tube, right and left ovaries were healthy and there were no pelvic adhesions.

A right total salpingectomy was performed. The patient made satisfactory post-operative progress. The post-operative PCV was 25% and skin sutures were removed on the fifth post-operative day. The histology report confirmed tubal ectopic gestation. She subsequently had an uneventful antenatal period and had spontaneous vaginal delivery of a live female baby weighing 3.2 kilograms (kg) on 30th of April 2009 with Apgar scores of 7 and 9 at one and five minutes respectively.

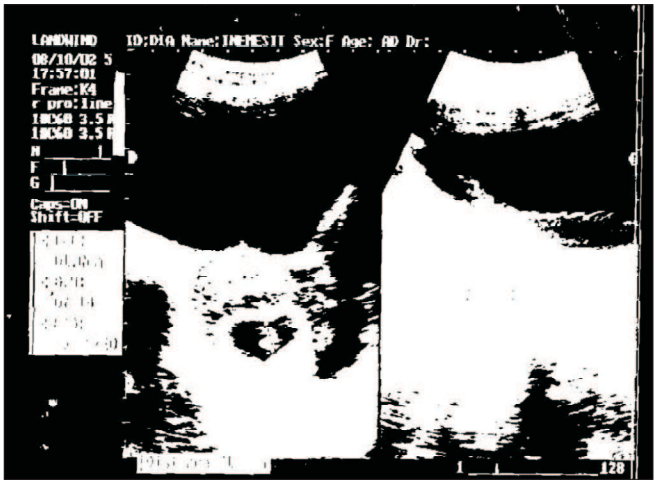

FIGURE I: A trans-abdominal ultrasound scan picture showing a bulky uterus with a gestational sac and a right adnexal mass.

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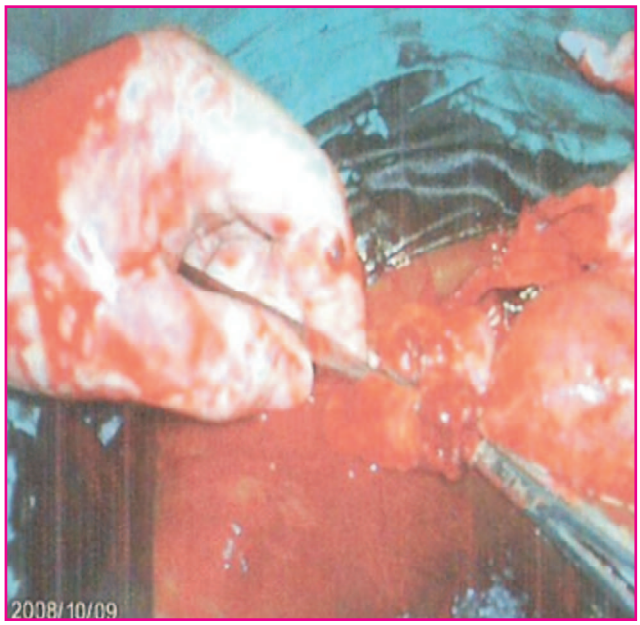

Discussion

Heterotopic pregnancy remains one of the greatest enigmas in gynaecological practice as no other pelvic condition is documented to give rise to more diagnostic errors [3]. The condition is potentially catastrophic for the mother and almost always fatal to the embryo or fetus when the ectopic component ruptures [4]. Its diagnosis is often difficult particularly in the absence of rupture as its associated symptoms are non-specific and may be attributed to complications of intrauterine pregnancy [2,4]. In addition, due to its rarity, it may be overlooked when an intrauterine pregnancy has been confirmed with ultrasonography [5]. Available evidence indicates that more than 50% of these pregnancies are identified by sonography or laparoscopy two weeks or more after the initial visualization of the intrauterine pregnancy though approximately 80% go undiagnosed before rupture of the ectopic component [6]. In majority of cases, the extrauterine component is tubal. However, cervical, ovarian, caesarean scar and abdominal heterotrophic pregnancies have all been reported [7,8].

Its incidence in spontaneous cycles was traditionally estimated to be about 1 in 30,000 pregnancies [2]. However, over the last three decades due to the increasing incidence of pelvic inflammatory disease, use of intrauterine contraceptive devices, tubal surgery and pharmacologic ovulation induction, its incidence has increased such that rates of I in 4,000 to 1 in 7,000 pregnancies are being reported [5,8]. Following ART the rate of heterotrophic pregnancy is increased more than

100 fold to about 1 to 3 % [7]. This is largely due to the presence of multiple embryos in a given cycle and the increased risk of tubal damage in an infertile population.

Interestingly, our patient had no risk factors for ectopic pregnancy and spontaneously conceived. She presented with amenorrhoea, abdominal pains and an USS report showing an intrauterine pregnancy. However, there should always be a high index of suspicion as the signs and symptoms of heterotopic pregnancy are usually non-specific as shown in this case when the findings on clinical examination and the trans-abdominal USS features mimicked other conditions such as a haemorrhagic corpus luteum, ruptured corpus luteal cyst, ovarian inflammation and an appendiceal phegmoma[9]. The diagnosis of heterotrophic pregnancy was confirmed by a trans-vaginal USS which showed an adnexal mass with a fetal node within it. The improved resolution of the trans-vaginal USS enables earlier and more accurate diagnosis of heterotrophic pregnancy and has significantly resulted in a reduction in the mortality associated with it [8]. As in this case, the demonstration of a live embryo within the gestational sac in the adnexa remains the gold standard for the diagnosis of ectopic pregnancy [5].

The management of heterotrophic pregnancy can pose great challenge. Its aim is to terminate the extrauterine pregnancy while retaining a viable intrauterine pregnancy[10]. However, the state of the tubes, the condition of the patient, facilities available, expertise of the surgeon and viability of the intrauterine fetus should be taken into consideration when instituting management. Administration of potassium chloride, hyperosmolar glucose and hypertonic saline using laparoscopy or ultrasonography to selectively target the ectopic component has been described prior to rupture of the ectopic component and when the patient is haemodynamically stable[9]. Laparoscopic surgery (salpingotomy or salpingostomy) can also be performed prior to tubal rupture when there is no haemoperitoneun. However, both modalities of treatment carry the risk of persistent trophoblastic tissue that may result in tubal erosion with subsequent intraperitoneal haemorrhage [12]. In addition, the effects of locally administered drugs on the developing foetus are not yet known and have not been studied [4]. When there is obvious tubal rupture with haemoperitoneum as in our case, laparotomy with salpingectomy is the safest option and gives the best results[10,13]. During surgery however, minimal handling of the uterus is essential to reduce reactionary undesired uterine contractions [9]. Speed in achieving haemostasis is also important as bleeding from uterine vessels might produce hypotension and reduce blood flow to the intrauterine fetus causing acute intrauterine asphyxia [9].

After the surgery our patient carried her pregnancy to term and delivered a live female baby. Available evidence indicates that 2/3rd of intrauterine pregnancies are delivered alive while 1/3rd are aborted [13]. However, in a comparison study of assisted versus (vs) spontaneous heterotopic conceptions, the assisted conception group had a higher live birth rates than the spontaneous group (47.8% vs 20%) [11].

In conclusion, spontaneous heterotopic pregnancy a rare but potentially fatal condition can occur in our environment. Clinicians should always maintain a high index of suspicion particularly in patients presenting with amenorrhoea and abdominal pains even when an intrauterine pregnancy has been confirmed and a thorough assessment of the adnexa using a trans-vaginal USS should be routinely performed. Early diagnosis and prompt intervention resulted in a good outcome with the subsequent delivery of a live baby at term in our patient.

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