Case Report

Live Birth after Treatment of a Ruptured Ectopic in a Spontaneous Heterotopic Pregnancy

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Abstract

Heterotopic pregnancy, though more common in pregnancies achieved by assisted reproduction, is extremely rare following natural conception. Owing to this rarity, the diagnosis is oftentimes missed or made late, usually following rupture of the extrauterine gestation. Heterotopic pregnancy, therefore, expectedly carries significant maternal mortality and morbidity. With the advancement in assisted reproductive techniques, and improvement in the diagnosis of ectopics using transvaginal ultrasound and beta-human chorionic gonadotropin assay, the frequency of heterotopic pregnancy is likely to increase, especially so in a country like Nigeria, with high twinning and ectopic pregnancy rates. To promptly diagnose heterotopic pregnancy and timely intervene, a high level of clinical suspicion is required. This would mitigate maternal morbidity and mortality, as well as preserve and continue the intrauterine pregnancy. With early diagnosis and prompt intervention, intrauterine pregnancy has a favourable outcome in the majority of cases. We present a 37-year-old multigravida with naturally conceived heterotopic pregnancy, in which the ectopic pregnancy ruptured. She subsequently had spontaneous vaginal delivery following surgical management (laparotomy) of the ruptured ectopic pregnancy.

Keywords: Ectopic pregnancy, heterotopic pregnancy, laparotomy, natural conception, salpingectomy

Introduction

Heterotopic pregnancy is one in which intrauterine and ectopic pregnancy occur concomitantly.[1] More common after assisted reproduction, heterotopic pregnancy very rarely occurs spontaneously, seen in only 1/30,000 natural conceptions.[1,2] The frequency of ectopic and heterotopic pregnancy is however rising globally, owing to a combination of increased utilization of assisted reproductive techniques (ART), and increased availability and advancement in transvaginal ultrasound scan and quantitative serum beta-human chorionic gonadotropin assay for early diagnosis.[3] Nigeria, aside from the having the highest twinning incidence worldwide, also has a high frequency of ectopic pregnancy.[4] A high rate of heterotopic pregnancy might therefore not be unexpected in Nigeria.[4]

This report is a case of ruptured spontaneously conceived heterotopic pregnancy. A laparotomy and right total salpingectomy were performed emergently, and the patient subsequently had a spontaneous vaginal live birth of the intrauterine gestation.

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Case Report

Mrs. AB, a 37-year-old gravida 3, para 2, with two living children, had initially presented to a private hospital in Osun state, in hypovolemic shock and symptoms suggestive of a ruptured ectopic gestation, following 10 weeks of amenorrhea. Ultrasound evaluation revealed a heterotopic pregnancy-an intrauterine gestation coexisting with a ruptured right tubal ectopic pregnancy. She had emergency exploratory laparotomy and right total salpingectomy, with intraoperative findings of a ruptured right fimbrial ectopic gestation, with two litres of haemoperitoneum, grossly normal contralateral fallopian tube

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and ovaries bilaterally, and a bulky uterus. The live intrauterine gestation was left *in situ*. She was transfused intraoperatively.

Pregnancy was spontaneously conceived. She had no prior ectopic pregnancy, tubal pathology, tubal or pelvic surgery. There was no history of multiple sexual partners or pelvic inflammatory disease (PID). She had used combined oral contraceptive pills before index gestation but had never used progesterone-only or intrauterine contraceptive devices. She did not smoke.

Following successful management of the ruptured ectopic, she received antenatal care in our facility. Booking parameters and investigations were all within normal limits. An ultrasound scan revealed a single viable intrauterine gestation. Her antenatal period was uneventful. At 34 weeks' gestational age, she presented in the active phase of labour. She was administered steroids for foetal lung maturation, following which she progressed in labour to spontaneous vaginal delivery of a live male baby, who weighed 2250 g at birth, with one and five minutes Apgar scores of 8 and 10 respectively. Both patient and her baby are currently doing well.

Discussion

Duverney discovered the first reported case of spontaneously conceived heterotopic pregnancy at autopsy in 1708.[5] Spontaneous heterotopic pregnancy occurs rarely, with a reported frequency of 0.08%. The use of ART increases this incidence to 1%.[6] As was the case in our patient, the ectopic site in a heterotopic pregnancy is frequently the fallopian tube, with the ampulla being the most common location, and the fimbria, which was the site in our patient, the least common.[5,7] Most heterotopic pregnancies reported are singleton intrauterine gestations,[2] just as in our patient.

Predisposing factors to heterotopic pregnancy are the same as for an ectopic, and these include a prior ectopic, PID, tubal surgery, use of ART and ovulation induction agents, among others.[1,8] Our patient had no identifiable risk factor. In patients like ours with no obvious risk factor, a very high degree of clinical suspicion is required to diagnose heterotopic pregnancy.

Being rare, heterotopic pregnancy is often unsuspected, leading to mis- and late diagnosis, and consequently, high morbidity and mortality.[4] Patients frequently present acutely with the ectopic rupture before the diagnosis is made,[8] as was in this case. Salvaging the intrauterine pregnancy and averting maternal mortality and morbidity requires timely diagnosis and prompt intervention.[9,10] In the reported case, though the diagnosis was not made until the ectopic rupture, a prompt intervention was very key in mitigating maternal morbidity and mortality, and preserving the intrauterine gestation.

Termination of the ectopic with preservation of the intrauterine gestation is the primary goal of management of heterotopic pregnancy.[8] This was successfully achieved in our patient. Surgical treatment options for tubal heterotopic pregnancy include laparoscopy or laparotomy. Laparoscopy is preferred, because asides from its well-established advantages over open surgery, it carries a lower risk of loss of intrauterine gestation, as it causes less irritation of the uterus at the surgery.[2,4] Laparoscopy is, however, not well established in resource-poor settings like Nigeria.[4] Most heterotopic pregnancies published thus far in Nigeria, have therefore been managed by laparotomy.[4] Laparotomy is also indicated when as in our case, the ectopic pregnancy is ruptured, with a large haemoperitonium, and the patient is haemodynamically unstable.[9] A favourable outcome for the intrauterine pregnancy, as was in this case, is reported in 50%–60% of cases.[8]

In conclusion, heterotopic pregnancy, though very rarely, can follow a spontaneous conception. Preventing maternal morbidity and mortality, preserving and continuing the intrauterine gestation, require a very high degree of clinical suspicion for timely diagnosis and prompt intervention. Timely intervention and proper management would result in a favourable outcome of the intrauterine pregnancy, as seen in the reported case.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has given her consent for her images and other clinical information to be reported in the journal. The patients understand that her name and initials will not be published and due efforts will be made to conceal her identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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