LIVE BIRTH AFTER LAPAROTOMY FOR RUPTURED HETEROTOPIC CORNUAL AND INTRAUTERINE TWIN GESTATION IN A SPONTANEOUS CYCLE: A CASE REPORT AND LITERATURE REVIEW.

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ABSTRACT

BACKGROUND: Heterotopic pregnancy is the coexistence of an intrauterine and extrauterine gestation. It is a rare occurrence especially in a natural conception. Its incidence is however increasing with the advent of assisted reproductive techniques.

CASE PRESENTATION: A rare case of triplet heterotopic cornual gestation involving twin intrauterine and a cornual gestation in a spontaneous conception. She presented with an acute abdomen and she had laparotomy with cornual resection. The intrauterine pregnancy remained uneventful till term.

CONCLUSION: Heterotopic pregnancy is a life threatening condition. Management with laparotomy can result in the survival of the intrauterine gestation till term without adverse outcome.

KEYWORDS: Heterotopic gestation, cornual ectopic gestation, term gestation.

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INTRODUCTION

Heterotopic pregnancy is the coexistence of intrauterine and extra-uterine gestation.[1] It is a rare occurrence with an estimated incidence in spontaneous conception of about 1 in 30,000 but as high as 1 in 3,600 to 1 in 100 in patients who had assisted reproductive techniques (ART) like in-vitro fertilization and embryo transfer (IVF-ET).[2] The incidence of cornual heterotopic pregnancy in spontaneous pregnancies is unknown[2] but an incidence of 1/3,600 has been reported in IVF pregnancies.[3] Nigeria has one of the highest rate of twin gestation worldwide and a high incidence of ectopic pregnancy, therefore, a high incidence of heterotopic gestation may be found in the country.[4]

Due to the rarity of heterotopic pregnancy, its diagnosis requires a high index of suspicion and the management requires careful interventions aimed at removing the ectopic gestation while maintaining the intrauterine gestation.

We present a case of ruptured heterotopic cornual gestation and intrauterine twin gestation with demise of one of the twins in a spontaneous conception. The woman had laparotomy with cornual resection of the ectopic component while the surviving twin was managed till term and was delivered.

CASE REPORT

The patient was a 40 year old unbooked $G_4P_2^{+1}$(2 Alive) at 10 weeks and 1 day of amenorrhoea from her last normal menstrual period. She presented at the emergency department with recurrent bleeding par vaginam of 3 weeks and recurrent lower abdominal pain of 2 week duration. Pregnancy was spontaneously conceived. She had a spontaneous incomplete miscarriage after a 6 week period of amenorrhoea 2 years prior to presentation for which she had a manual vacuum aspiration done with no post-abortal sequelae.

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She had no history of pelvic inflammatory disease or ectopic pregnancy.

On examination, she was pale and had a blood pressure of 80/60mmHg and pulse rate of 120bpm, thready and of low volume. There was generalised abdominal tenderness with guarding and rebound tenderness. There was no active bleeding. The cervix was smeared with altered blood. There was bilateral cervical motion tenderness and the pouch of Douglas was boggy.

Her haemoglobin level was 6.3 g/dl and a transabdominal ultrasound scan revealed two foetuses at the fundal region with a dividing membrane separating them. One of the foetuses had cardiac activity and a crown-rump length (CRL) of 3.36cm (corresponding to gestation of 10weeks +5days) and the other had no cardiac activity and a CRL of 3.2cm (corresponding to gestation of 9weeks+6days). There was large amount of fluid in the peritoneum. There was no mention of any gestation outside the uterine cavity.

A diagnosis of acute abdomen with ruptured heterotopic gestation was suspected due to both the clinical, haematological and ultrasound findings of intrauterine gestation with large amount of peritoneal fluid. She was resuscitated with intravenous crystalloids and counseled for emergency exploratory laparotomy.

At laparotomy, we found a 16 week size uterus with ruptured right cornual ectopic gestation with about 2cm extention to the body of the uterus as an incomplete rupture with broad ligament haematoma. There was also about 2.5litres of haemoperitoneum. Right cornual resection and uterine repair was done in two layers with Vicryl 2 and 2/0 carefully (figure 1). Haemostasis was secured. The broad ligament haematoma (about 800mls) and haemoperitoneum were evacuated and the peritoneum was lavaged. She had two units of blood transfused intra-operatively and another unit after the surgery.

She was placed on Nifedipine and Dydrogesterone tablets both at 10mg twice daily for two weeks. The postoperative hemoglobin level was 10.3g/L and the histopathological examination of the excised cornual and right fallopian tube confirmed an ectopic pregnancy.

She was discharged on the 14th day after surgery to booking clinic where she was seen and scheduled for an elective caesarean section at 38weeks. Ultrasound done at four weeks after the surgery showed a single viable intrauterine pregnancy at 14weeks and 6 days. Her antenatal period was uneventful and her pregnancy continued till 37weeks and 2 days when she had an emergency caesarean section when she presented with labour pains. Intraoperatively, the right cornual end was observed to have healed well (figures 2 and 3). She was eventually delivered of a live and healthy female baby with birth weight of 2.66kg and an Apgar scores of 8 and 10 at the first and fifth minutes respectively.

DISCUSSION

Heterotopic gestation is a very rare condition in spontaneous conception with an incidence of 1 in 30,000 in the general population but this has greatly increased with the advent of ART to an incidence of 1 in 100.[2] Cornual heterotopic gestation is even less common compared to other types of tubal heterotopic gestation. It occurs in 1/3600 IVF-ET pregnancies.[3] About 75% of reported cornual heterotopic pregnancies occur in IVF-ET conception while 16.7% occur in spontaneous conception and 8.3% in superovulation cycles.[3]

The risk factors for heterotopic gestation are the same as that of ectopic gestation.[5] These include pelvic infection, tubal surgery, previous ectopic pregnancy and ART.[1,5] Heterotopic gestation may however occur without any risk factor.[3] In this patient, the pregnancy was spontaneously conceived and had no identifiable risk factor.

The commonest presentation is abdominal pain.[6,7] Other symptoms include vaginal bleeding, acute abdomen and shock.[5,6,7,8] Abdominal pain and vaginal bleeding were the symptoms in this patient. Up to 41.2% of patients with cornual heterotopic pregnancy may however be asymptomatic especially when unruptured.[3]

While there is a delay in the diagnosis in about 33% of all heterotopic gestation[9], only about 10% of cornual heterotopic gestation is diagnosed before surgery.[10]

The accuracy of the diagnosis of heterotopic gestation can however be enhanced with the aid of transvaginal ultrasound (TVS). Studies have reported accurate diagnosis in 71.45% to 92.4% of cases.[11,12] This wide variation may be due to the gestational ages (GA) at which the TVS was done. Detection rate of heterotopic gestation by TVS seems to reduce as GA advances with a rate of 70-72% at 5 weeks and 10% at GA >11weeks.[9,12] Another reason may be due to the detection of an intra-uterine gestation on ultrasound which may give a false assurance that precludes the examination of the adnexae for co-existing extra-uterine gestation.

Mortality rate among women with heterotopic gestation is 1%.6,13 The mean age of rupture for

cornual heterotopic gestation is 12.1weeks.[3] Rupture in this patient occurred at 10.1weeks. Beck et al[14] have however reported a case of ruptured cornual heterotopic pregnancy at 26 weeks with a live fetus. The extra-uterine component may however remain unruptured till the intrauterine component reaches term as reported by Ogunniyi et al[15] in a case of obstructed labour caused by an advanced heterotopic Ovarian pregnancy.

Transabdominal ultrasound done for our patient did not detect the cornual ectopic gestation. This may be due to the fundal location of the intrauterine twin gestation which could have led to the overcrowding of the fundal region. But the diagnosis of heterotopic gestation was made based on the history and clinical findings of an acute abdomen with ultrasound findings of an intrauterine gestation and large amount of peritoneal fluid in the patient.

The management of heterotopic gestation poses a great challenge due to the presence of an ongoing intrauterine gestation. The options of treatment for heterotopic gestation are medical and surgical. Compared to medical intervention, surgical management has a higher live birth rate (60.9% vs 50%) and lower miscarriage rate (13% vs 50%).[3] The surgical option may be laparoscopy or laparotomy.[3] Expectant management, with the hope of spontaneous resolution of the ectopic component, is not encouraged due to lack of guidelines for this form of management and risk of rupture.[16]

Medical management is an option in haemodynamically stable patient with unruptured ectopic component and a beta-hCG level lower than 1000 mIU/ml.[3] Drugs that can be used are potassium chloride and hyperosmolar glucose.[3,7,16] Methotrexate and prostaglandins are contraindicated because of their adverse effect on the viable intrauterine gestation. Routes of administration include laparoscopy or ultrasound guided for viable co-existing intrauterine gestation and hysteroscopic and systemic routes for non-viable intrauterine gestation.[2,16]

Surgery, either via laparoscopy or laparotomy, is the primary mode of management for heterotopic pregnancies.16 Laparoscopy is more common and preferred to laparotomy except in cases of haemodynamic instability with intraperitoneal hemorrhage where laparotomy may be the better option of treatment,8,16 as was done for this patient. Conversion to laparotomy however occurs in approximately 27% of laparoscopic cases due to haemoperitoneum or technical difficulties.[2] Laparotomy may also be the only surgical option available especially in a low resource setting. In Nigeria, laparotomy is the most common surgical treatment for heterotopic gestation.[17,18] Badejoko and colleagues however reported a case managed with laparoscopic salpingectomy.[4] The surgical procedures that can be done for cornual heterotopic gestation include cornual resection and placement of vicryl loop,[16] while salpingectomy can be done for other forms of tubal ectopic.[19]

Generally, the live birth rate for the intrauterine fetal component of heterotopic gestation after treatment is between 66.2% and 70.5%[12,13] but it is between 64.5% and 57.6% for cornual heterotopic gestation if treatment is given before and after onset of symptoms respectively.[3] The intrauterine gestation of this patient was carried to term and the baby was delivered via caesarean section without any obstetric complication.

CONCLUSION

Heterotopic cornual with an intrauterine twin gestation in a spontaneous cycle is a very rare occurrence. To the best of our knowledge, this is the first reported case of a delivery of a viable fetus after laparotomy for a cornual heterotopic pregnancy.

Early diagnosis and treatment have a good prognosis for both the woman and the intrauterine gestation. The presence of intrauterine gestation should not preclude the assessment of the adnexae for extra-uterine gestation.

Laparotomy with cornual resection can be done when necessary and the survival of the intrauterine gestation till term is feasible without adverse outcome. This is especially important in a low resource setting where access to laparoscopy may be limited.

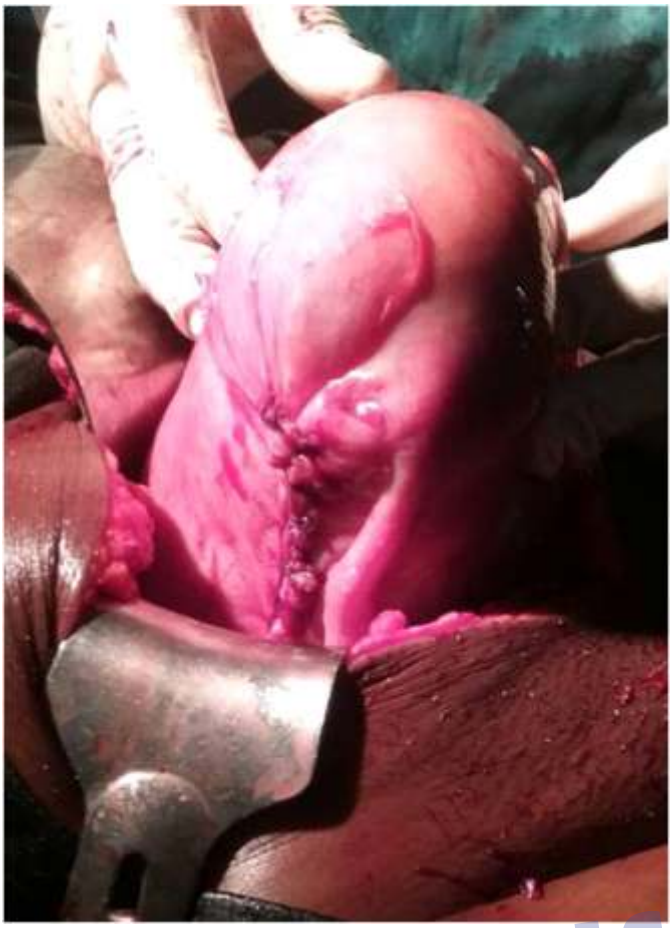

Figure 1: The gravid uterus after right cornual resection and evacuation of broad ligament haematoma

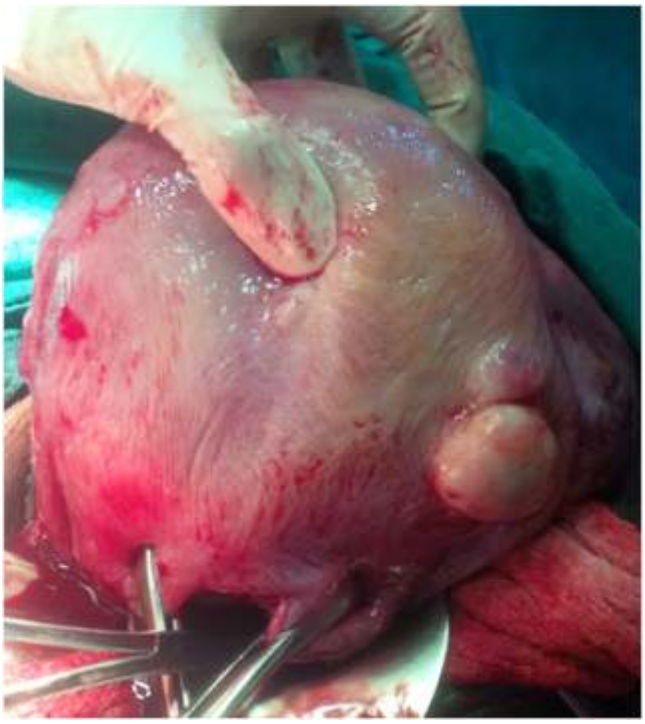

Figure 2: The uterus at caesarean section

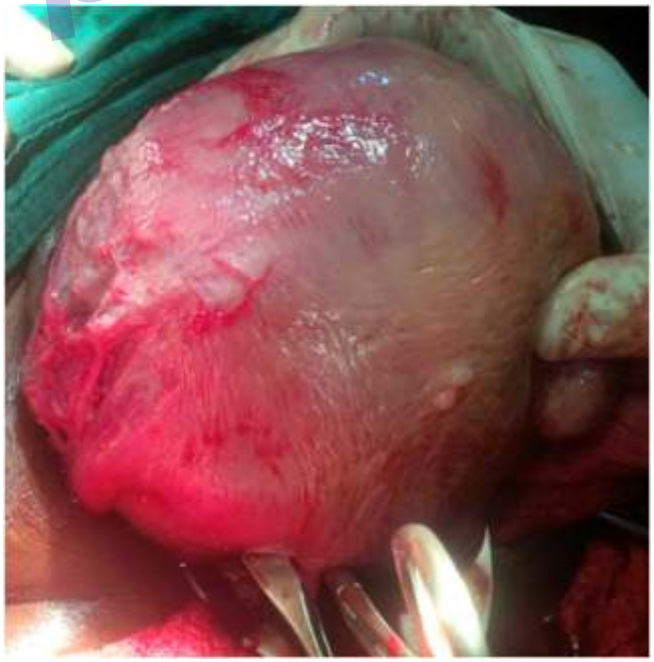

Figure 3: The well healed right cornual end of the uterus at caesarean section.

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