

Misdiagnosed ruptured cornual ectopic pregnancy: Case report and literature review

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Abstract

Ruptured ectopic pregnancy is a leading cause of maternal morbidity and mortality worldwide. Cornual ectopic pregnancy is a specific type that occurs at the horn of the uterus. It often ruptures later than the more common tubal ectopic pregnancies because of the more distensible myometrial tissues surrounding the gestational sac. The diagnosis of cornual pregnancy poses a lot of challenges to sonographers because of its closeness to the body of the uterus necessitating a lot of skill and expertise. Rupture of a cornual ectopic pregnancy is life threatening because of associated massive haemorrhage. Treatment is mostly by emergency laparotomy and cornual resection with salpingectomy or hysterectomy.

Here, we report a case of cornual ectopic pregnancy at 11 weeks gestational age that was misdiagnosed as intra uterine gestation until it ruptured. Patient had laparotomy with right cornual resection and ipsilateral salpingectomy. Ruptured cornual ectopic pregnancy and its morbidities and mortalities should be prevented through detailed early first trimester examinations. Ultrasonography especially the transvaginal modality remains a very helpful but occasionally inconclusive tool in the diagnosis of this life-threatening condition.

Keywords: Misdiagnosis; Cornual; Ectopic; Pregnancy

1. Introduction

Interstitial and cornual ectopic pregnancies are rare but are the most dangerous forms of ectopic pregnancies accounting for 2-4% of all ectopic pregnancies¹. An interstitial pregnancy is defined as a gestational sac that implants within the proximal intramural portion of the fallopian tube that is enveloped by the myometrium^{2,3}. Interstitial pregnancy is often used interchangeably with cornual pregnancy and this is applicable to this report. The commonest site of ectopic gestation is the ampullary region, followed by the isthmus, fimbriae, cornual and tubo-ovarian regions.⁴ Most cornual ectopic pregnancies present in ruptured state⁴. Because of the ability of the myometrial tissues surrounding the ectopic gestational sac to stretch to variable degrees, a cornual rupture generally occurs after 9 weeks and as late as 20weeks⁵. Rupture of the gestational sac at such an advance age is often associated with catastrophic haemorrhage and up to 2.0-2.5% maternal mortality rate which is 6 – 7 times higher than that in other forms of ectopic pregnancies^{1,6}. About 4 out of 11 deaths from ectopic pregnancies result from cornual ruptures because haemorrhagic shock is found in almost a quarter of the patients⁶. This is due in part to delay in diagnosis as well as the speed of haemorrhage accompanying the rupture⁷.

The diagnosis of interstitial pregnancy is very difficult because the classical triad of ectopic pregnancy - amenorrhoea, abdominal pain, and vaginal bleeding occur in less than 40% of the patients⁸. There is low sensitivity and specificity of symptoms and imaging due to the proximity between the implantation site, accompanying intrauterine invasion and the endometrial cavity⁸. The diagnostic criteria for cornual ectopic pregnancy include ultrasound findings of an empty

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uterus, a gestational sac located eccentrically and more than 1cm from the most lateral wall on the uterine cavity, and a thin (less 5mm) myometrial layer surrounding the gestational sac^{8,9}.

The treatment of ruptured interstitial/cornual pregnancy is traditionally by surgical cornual resection mostly through laparotomy. Sometimes, a hysterectomy is may be indicated due to the haemorrhage¹⁰. Currently, in unruptured cornual ectopic with asymptomatic presentation, a more conservative laparoscopic treatment or even medical treatment can be accomplished with great success and with less unfavourable effects on future pregnancies¹¹.

2. Case report

Mrs O.C was a 36-year-old business woman, now Para 2⁺¹ (2 Alive) who resided at New Haven, Enugu. She was a Christian of the Roman Catholic denomination and an Igbo from Enugu state.

She was referred from a private hospital on account of persistent abdominal pain of 3 days duration and deteriorating haemodynamic status after 11weeks gestational age. She was rushed to the private hospital on account of sudden onset of severe abdominal pain of 3 hours duration. Pain started at the right iliac fossa and later became generalized. Her referral letter revealed that she was in painful distress, afebrile, anicteric, and pale while her chest was clinically clear. Her vital signs on admission showed a pulse rate of 80 b/min. B.P = 108/67mmHg with a temperature of 36.3° C. Her abdomen was full and moved with respiration. There was a sub-umbilical midline scar and generalized tenderness restricting deep palpation. She vomited three times while being examined at the hospital.

She was given intravenous injections of pentazocine 30mg, omeprazole 40mg, paracetamol 600mg, promethazine 25mg and intravenous infusion of 0.9% Normal Saline before sending her for abdominopelvic scan (Figure 1) which 'revealed a live in-utero fetus at 11weeks and 2days gestational age with no evidence of haemoperitoneum, ectopic gestation, or submembranous bleeding'.

From the above report patient was admitted at the hospital and managed conservatively on the above medications without a clear diagnosis until patient's vital signs began to deteriorate. Her pulse rate rose to 95 b/min while the B.P dropped to 75/55mmHg with patient becoming increasingly dizzy with increasing abdominal pain. For this reason, she was referred to Enugu State University Teaching Hospital (ESUTH) Parklane, Enugu but relatives decided to rush patient to Hope Hospital and Maternity for prompt expert management. She spent about 16 hours at the hospital before she was referred.

She had two spontaneous vaginal deliveries at term in the past 5 years of her marriage without any complications. Her last child was 3 years old and her husband resided in Dubai, United Arab Emirate. She had ovarian cystectomy at a private hospital 12 months prior to conceiving this pregnancy without complications and there was no previous history of pelvic infections.

On presentation patient was found to be conscious and alert but in shock. She was afebrile, anicteric, pale (urgent PCV= 15%) with cold clammy extremities. Her pulse rate was 140 b/min, low volume, fast and thready. Her B.P = 50/? mmHg. The abdomen was distended and severely tender with guarding. Urgent bedside ultrasound revealed massive haemoperitoneum with an intact gestational sac containing a fetus with no cardiac activity floating in the peritoneal cavity. The fetus corresponded to 11 weeks gestational age. The uterus was bulky and the endometrial cavity was empty. A diagnosis of ruptured ectopic pregnancy possibly an abdominal pregnancy was made and the patient and her relatives were counseled and informed consent obtained for emergency exploratory laparotomy. Immediate resuscitative measures were commenced. A second intravenous access was secured with size 16 canular and infusion of crystalloids maintained. Samples were collected for grouping and crossmatching of 4 units of blood and for other investigations. Indwelling Foley catheter was passed. Intra-nasal oxygen was commenced at 6L/min.

She had exploratory laparotomy under general anaesthesia and the findings showed

- -Massive haemoperitoneum approx 2.5litres.
- -Extruded 12-week-size fetus covered with intact fetal membranes and floating in clear liquor amnii (Figures 2 and 3).
- -Ruptured right uterine cornua with friable surfaces.
- -Intra-peritoneal adhesion bands involving the right fallopian tube, the uterine body and the pelvic side wall.
- -Both ovaries and the left fallopian tube appeared grossly healthy.

Right cornual resection with right salpingectomy was performed and repaired with Vicryl-2 (R) sutures. Haemostasis was secured. The haemoperitonium was evacuated and the peritoneal cavity cleaned up. The anterior abdominal wall was closed in layers using Vicryl 2/0 (R) for the peritoneum, subcutaneous layer and for the skin (subcortically). The rectus sheath was closed with Vicryl-2 (R) and the wound site dressed with sterile gauze and plaster.

She was transfused a total of 5 units of whole blood and was discharged on the 4th post operative day in good clinical condition with a post transfusion PCV of 28% after due counseling. Her two-week post operative follow-up visit was satisfactory.

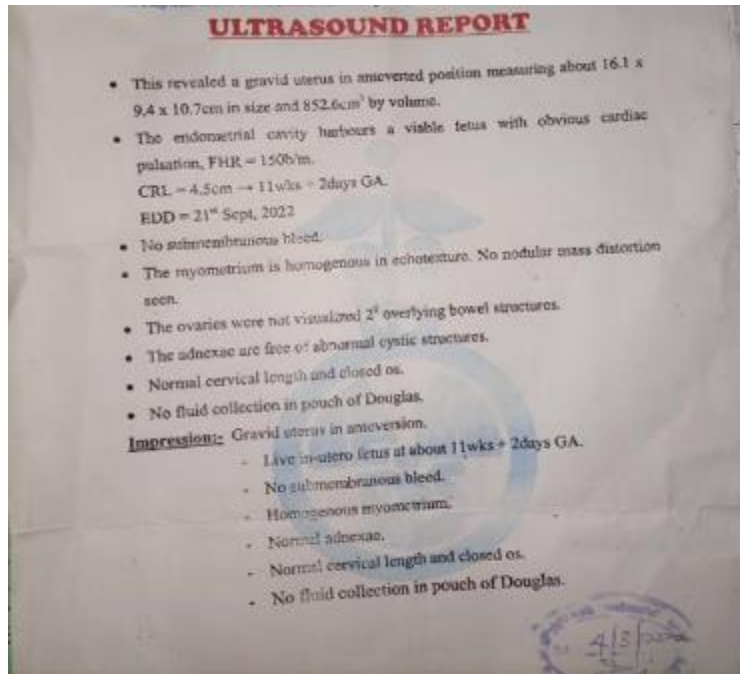


Figure 2 Ultrasound scan reported intrauterine gestation

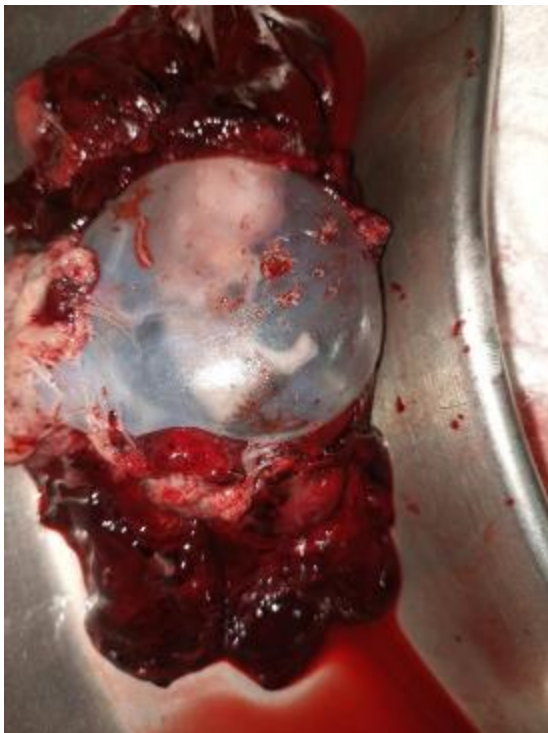


Figure 3 The products of conception with intact gestational sac



Figure 4 Parts of the uterus at surgery

3. Discussion

Missed diagnosis of cornual ectopic often result in catastrophic outcome as witnessed in the index case. Ruptured cornual ectopic pregnancy is as life threatening as its diagnosis is difficult⁷. The distensile nature of the myometrium surrounding the ectopic mass ensures that rupture occurs late often between 9-20 weeks^{1,5,10}. Mrs O.C ruptured at 11weeks gestational age. The commonest diagnostic pitfall is the tendency to mistake the cornual ectopic sac for intrauterine normal pregnancy occasioned by obvious myometrial surroundings and the proximity to the endometrial cavity. This was clearly the situation with the index case. Louis et al reported that of 36 women diagnosed with interstitial pregnancy, 15 (41.7%) were misdiagnosed with 14 of them mistaken for intrauterine pregnancy (both viable and nonviable). He reported that making the right diagnosis gets even more difficult as the gestational age increases despite advances in sonographic skills and equipment¹². Sant and Anderson reported a case of a misdiagnosed ruptured cornual pregnancy occurring at 21 weeks of gestation where both ultrasound examination and computer tomography had revealed no sign of abnormal pregnancy¹³.

The most feared consequence of misdiagnosed cornual ectopic pregnancy is uterine rupture leading to torrential haemorrhage, hypovolemic shock, and death^{1,6,7,10,13}. Our patient presented in shock with a pulse rate of 140 b/min and blood pressure of 50mmHg systolic and undetectable diastolic pressures after 16 hours of missed diagnosis and management. She was subjected to some needless treatments including multiple intravenous injections of pentazocine, omeprazole, paracetamol along with infusions of normal saline before ultrasound misdiagnosed the ectopic pregnancy as intrauterine gestation. It took the deterioration of her vital signs for referral to be affected. Louis et al also reported that 6 out of the 36 patients they studied had had uterine evacuations for suspected intrauterine nonviable pregnancies while in 2 of the cases the empty uterus was misdiagnosed as uterine fibroid coexisting with “intrauterine pregnancy”. Both later cases ruptured at 8 and 20weeks gestation respectively¹³.

The classical diagnostic criteria for cornual ectopic pregnancy by Timor-Tritsch et al include ultrasound finding of an empty uterus, a gestational sac located eccentrically and more than 1cm from the most lateral wall on the uterine cavity, and a thin (less 5mm) myometrial layer surrounding the gestational sac⁸ may not always be present because later studies showed that these criteria have 88-93% specificity and only 40% sensitivity^{8,13,14}. These limitations of ultrasound scan in diagnosing cornual ectopic pregnancies should always be remembered so that a high index of suspicion must be maintained.

The possible risk factors for cornual ectopic pregnancy in this patient include a previous history of pelvic surgery (ovarian cystectomy) performed 12 months earlier and, advancing maternal age. These may have resulted in proximal intratubal adhesions and increased risk of pelvic infections in the absence of other clear risk factors. Other predisposing risk factors for cornual pregnancy documented in literature include history of assisted reproductive techniques¹⁵, age >35 years^{16,17}, smoking¹⁸, history of pelvic inflammatory disease and sexually transmitted diseases, rudimentary uterine horn, previous ipsilateral salpingectomy or tubal injuries^{19,20}, failed use of intra uterine contraceptives^{18,21}, infertility²², previous pelvic surgeries and previous ectopic pregnancy^{4,23}. There may be cases with no detectable risk factors⁸.

The management of cornual pregnancies depends primarily on the hemodynamic status of the patient at presentation and, secondarily on the gestational age, fertility desires, available treatment facilities, and surgeon experience^{7,24}. Ruptured cornual ectopic pregnancy often accompanied with haemodynamic instability, is a surgical emergency. Immediate resuscitations followed by surgical cornual resection or cornuectomy is performed through the most expedient route usually laparotomy, but sometimes laparoscopically^{9,10,25} Surgery is both diagnostic and therapeutic⁴. This was the case with index patient who had emergency laparotomy, right-sided cornual resection and salpingectomy with good haemostatic control. Sometimes, hysterectomy may be indicated usually due to uncontrolled haemorrhage⁹. The aim of surgery is to rapidly secure haemostasis with adequate cornual reconstruction to prevent uterine rupture in future pregnancies. The integrity of the myometrium after cornual resection and reconstruction is unclear and so there is need to preserve normal uterine tissue, avoiding tissue damage by electrosurgery, and excising as minimal cornual tissues as possible^{26,27}. This will require good surgical approach and necessitate advanced laparoscopic skills and techniques^{25,28}. All possible measures should be put in place to reduce blood loss during surgery. These may include insertion of misoprostol vaginally, intra-myometrial injection of vasopressin, application of tourniquets and uterine devascularization²⁹⁻³².

When cornual ectopic pregnancy is detected unruptured, conservative management approaches may be indicated. These may involve such procedures as cornuostomy instead of cornual resection or cornuectomy, and laparoscopy in place of laparotomy⁷. Many cases of laparoscopic cornuostomy have been reported in literature^{9,10,11} and was aimed at better preservation of the integrity of the uterus for future fertility.

When medical treatment is indicated, intra-amniotic injection of Potassium Chloride, intra-placental or systemic methotrexate is the method of choice^{7,33} especially when the gestational sac diameter is less than 5cm³⁴. Unfortunately, some studies showed that methotrexate treatment was associated with up to 9–65% failure rate^{35,36} that may lead to ruptured uterus³³.

The main complications of treated interstitial pregnancies are recurrence and uterine rupture in subsequent pregnancies. Recurrence is the prerogative of medical treatment and cornuostomy, whereas uterine rupture is secondary to surgical treatment by cornual resection, probably due to the fragility of the uterine wall.

It is mandatory that detailed counseling and close antenatal follow-up be given to patients with history of cornual ectopic pregnancy. Early transvaginal ultrasound should be performed 5–6 weeks after the last menstrual period to confirm correct implantation of the subsequent gestation³⁷. It is advisable to plan patients for elective caesarean section to reduce the risk of uterine rupture in labour^{28,37} since the integrity of the uterus at the point of cornual rupture cannot be guaranteed with certainty.

4. Conclusion

Misdiagnosis of cornual ectopic pregnancy is a time-bomb waiting to explode. Cornual rupture is often catastrophic due to torrential haemorrhage usually leading to hypovolemic shock and even death. Clinicians are encouraged to develop a high index of suspicion and to always “think ectopic” especially when the classical symptoms and signs are equivocal. Training and retraining in ultrasonography will help to improve skill and reduce the incidence of misdiagnosis of cornual ectopic pregnancy.

Compliance with ethical standards

Disclosure of conflict of interest

No conflict of interest to disclosed.

Statement of informed consent

Informed consent was obtained from all individuals and participants included in the study

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