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Ruptured Interstitial Ectopic Pregnancy at 16 Weeks of Gestation Resulted in Maternal Near-Misses: Report of Two Cases and Literature Review

Research Article

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Abstract

Background: Interstitial ectopic pregnancy is a rare type of ectopic pregnancy that commonly results in uterine rupture and life-threatening hemorrhage, including death. Delayed or missed diagnosis contributes to an increased incidence of poor maternal outcomes.

First case report: A 32-year-old multipara women at gestational age of 16 weeks came with a complaint of sudden abdominal pain accompanied by diarrhea and vomiting. On arrival, blood pressure was unrecordable and she had white paper conjunctivae. Ultrasonography revealed massive intraabdominal fluid collection and abdominally located nonviable fetus. Laparotomy performed, 3.5L of a blood was sacked, repair of large fundal defect was performed, and the postoperative time was uneventful.

Second case report: A 25-year-old grand multipara woman with four months of amenorrhea came with a history of abdominal pain and vaginal bleeding of three days duration. Hemodynamically unstable, her hemoglobin was 4 g/dL at arrival. Ultrasonography revealed massive intraabdominal fluid collection and abdominally located nonviable fetus. Subtotal hysterectomy was performed and her postoperative time was uneventful.

Conclusion: Delay in health-seeking behavior and deferral in intervention of interstitial ectopic pregnancy could be associated with grave complications. High index of suspicion, immediate exploratory laparotomy is crucial and lifesaving than delay of imaging studies to reach concrete diagnosis.

Keywords: Interstitial Ectopic Pregnancy, Maternal Death

Introduction

Interstitial ectopic pregnancy is defined by the implantation of the blastocyst in the interstitial part of the fallopian tube within the myometrium [1-3]. This proximal part of the tube is approximately 2cm long and 0.7 mm wide and is described as intrauterine, but lies outside the endometrial cavity [1,4]. Due to their close anatomical proximity, interstitial, cornual, and angular pregnancy are often used interchangeably, but they must be differentiated [4,5]. When the blastocyst implanted in either horn of a bicornuate uterus, or a rudimentary horn of a unicornuate uterus or in the lateral half of a septated or partially septated uterus is called cornual pregnancy while implantation that takes place medial to the utero-tubal junction and round ligament is angular ectopic pregnancy [1,4-7]. Due to the anastomosis of the ovarian artery and the uterine artery cornus of the uterus is highly vascularized and chorionic villi may invade the blood vessels which can lead torrential hemorrhage if ruptured [8].

Interstitial ectopic pregnancy is a rare form of ectopic pregnancy, with a reported incidence of 2–3% of all ectopic pregnancies and as-

sociated with substantial mortality, accounting for 20 % of all deaths related to ectopic pregnancies [4,5,8-11]. Predisposing factors associated with interstitial ectopic pregnancy include delay childbirth until their later years, pelvic surgery, previous ectopic pregnancy, assisted reproductive technology, malformed uterus, and pelvic inflammatory disease [3,8-10,12]. The classic presentation of ectopic pregnancy with amenorrhea (positive pregnancy test), pain in the abdomen, and vaginal bleeding is found in less than 40% of cases [1,6,12]. The clinical presentation of interstitial ectopic pregnancy is not uniform and patients may develop severe complications and death unrecognized [12]. Symptoms occur 9-12weeks after the LMP and cervical excitation is not a specific sign [10]. Regular and quality antenatal care with perfect ultrasounds location can help in early diagnosis thereby preventing complications associated with ectopic pregnancy [12].

The diagnosis of interstitial pregnancy is challenging and often leads to delayed diagnosis and intervention, contributing to a higher mortality rate [5]. Ultrasonography diagnostic criteria for these type of ectopic pregnancies includes: empty uterine cavity, a separate gestational sac and at least 1 cm from the lateral edge of the uterine cavity and a thin (<5mm) myometrial layer surrounding the chorionic sack. The presence of the interstitial line sign, a line that extends from the upper region of the uterine horn to border to border of the intramural portion of the fallopian tube, also adds to the diagnosis [5,6,9]. Magnetic resonance imaging may be an important tool, particularly in cases of suspected second trimester rupture when ultrasound findings become more equivocal [5].

Management plan of interstitial ectopic pregnancy should individualized considering the gestational age of the ectopic pregnancy at time of diagnosis and the need for preservation of fertility but split into medical and surgical management [2,7-9]. Medical management consists of single-dose or multi-dose of local or systemic methotrexate injection with reported success rates between 66% and 100% [9]. Early gestation, and diameter less than 4 mm and serum ß-hCG levels less than 10.000 IU/L are essential to select medical treatment in an interstitial pregnancy [9]. Cornual wedge resection remains the best choice of surgical management of interstitial pregnancy to handle the bleeding, especially in haemodynamically non-stable patients but hysterectomy can be also performed [6,9].

Case Report 1

This is a 32-year-old Eritrean woman brought to Orotta National Referral Maternity Hospital with a complaint of sudden onset of severe lower abdominal pain, three episodes of profuse watery diarrhea, and vomiting of 3 hours period. She experienced mild lower abdominal pain one day before the current presentation, which subsides on its own without analgesia. She had a history of amenorrhea of four months and was not aware of her pregnancy. She denied having a history of vaginal bleeding, trauma, or drug intake. She had a history of cesarean delivery for fetal distress in her last pregnancy.

On arrival, she was in hypovolemic shock, drowsy not able to support her body with cold and pale extremities. Her vital signs were un-recordable blood pressure, pulse rate 127b/m, respiratory rate of 35 breaths/minute and Spo2 91% at room air. She had paper white conjunctivae and non-icteric sclera. Abdomen was moderately distended with diffuse tenderness and rebound tenderness. The fundal height was not clearly identified and on per vaginal examination cervical OS was tightly closed without blood in the examining finger.

With an impression of hypovolemic shock, two large-bore cannulas were secured and blood was drawn for complete blood count (CBC) and x-match. Transabdominal USG showed massive intra-abdominal fluid collection, empty uterus, nonviable fetus located outside the uterus, and distorted uterine fundus on the left side. (Figure: 1) The CBC result showed hemoglobin of 9g/dl and the pregnancy test was positive. Four litters of crystalloids were given within one hour and the first O-ve blood arrived, which raised BP to 80/40 mmHg. With the final diagnosis of hypovolemic shock secondary to uterine rupture a high risk consent for possible hysterectomy was obtained and 2 grams prophylactic antibiotics, ceftriaxone was given.

Under general anesthesia and aseptic technique, midline incision was made. After entering the peritoneum, 3.5L of blood was sucked and the fetus with the placenta was removed from the abdominal cavity. The left uterine funds was found to be ruptured and the placental remnants at the rupture site was manually removed. (Figure 2) Repair of the rupture with number 1 delayed absorbable stich was performed in three layers and hemostasis secured. Intraoperatively, she was transfused with two unit of blood. After full instrument and gauze count, the abdomen was closed layer by layer. Postoperatively, another three units of blood, three fresh frozen plasmas given and her hemoglobin was 11.5g/dl during discharge. With uneventful postoperative time, she was discharged after 4 days with follow-up every two weeks for serial beta HCG level monitoring. During her follow-up the beta HCG weans out in 5 weeks and the family advised about contraceptives and they preferred the implant which was inserted during her last follow up.

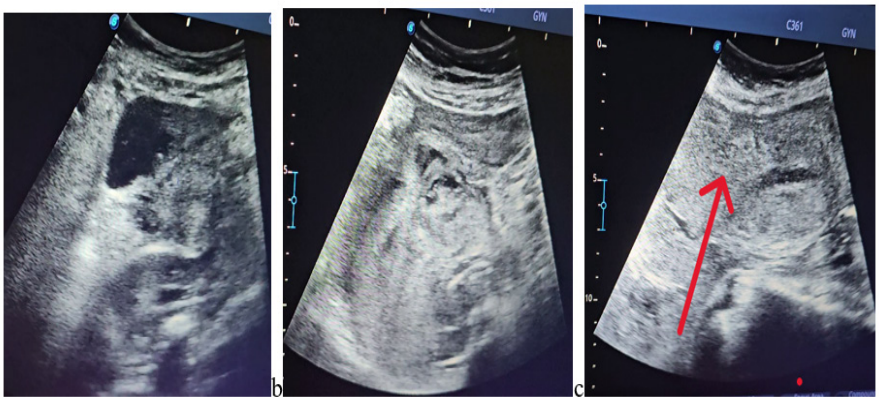

Figure 1: Ultrasonography results (a) showing massive intra-abdominal fluid collection (b) big intra-abdominal blood clots signifying rapid accumulation of blood (c) empty uterus with fundal defect (red arrow) continued with big clots.

Case Report 2

This is a 25-year-old G4P3003 Eritrean women admitted to Orotta National Referral Maternity Hospital as a case of ruptured ectopic pregnancy. She didn't remember her last menstrual period, but she claims to be amenorrhic for four-month period. On presentation, she was complaining of generalized abdominal pain and vaginal bleeding of three days. Later it was associated with abdominal distention, nausea, dizziness, and shortness of breath. She has never had any gynecological, obstetric, medical, or surgical problems before.

On physical examination: the patient was in respiratory distress and

her vital signs were a blood pressure of 70/40mmHg, a pulse rate of 110 beats / minute, a respiratory rate of 36 breaths/minute, SPO2 90% in room air and temperature of 36.5 °C. The abdomen was moderately distended with generalized tenderness and rebound tenderness. The uterus was palpable at 14-16 weeks of fundal height and on vaginal examination cervix was closed with tenderness of the cervical movement, blood on examining figure. Trans- abdominal ultrasound revealed massive intraperitoneal fluid collection, empty uterus with intact amniotic sac containing a nonviable fetus 16 weeks' gestation in the right lower quadrant of the abdomen. The complete blood count revealed hemoglobin of 4.16g/dl, WBC 20.57x103 per microliter with neutrophils 81.27% and platelet 336x103per microliter.

With the diagnosis of ruptured ectopic pregnancy and hemodynamic instability, she was resuscitated with crystalloids and transfused with two units of packed red blood cells preoperatively. The patient received 2 grams of ceftriaxone for prophylaxis and consented to high-risk laparotomy and possible hysterectomy. Under general anesthesia and aseptic technique an infra-umbilical midline incision was made. After opening the parietal peritoneum, 2.7 liters of blood were evacuated. The amniotic sac was found intact with the placenta floating in the peritoneal cavity. (Figure 3b) The uterus was exteriorized, which revealed a ruptured right uterine horn (Figure 3a,c). Subtotal hysterectomy was performed and both ovaries were reserved. Intraoperatively, she was transfused again with another two units of blood and the two fresh frozen plasma. Postoperatively, two units of blood, one fresh frozen plasma, were administered, and her hemoglobin increased to 12.5g/dl. The patient was discharged on the fifth postoperative day without any complications and the outpatient follow up was uneventful.

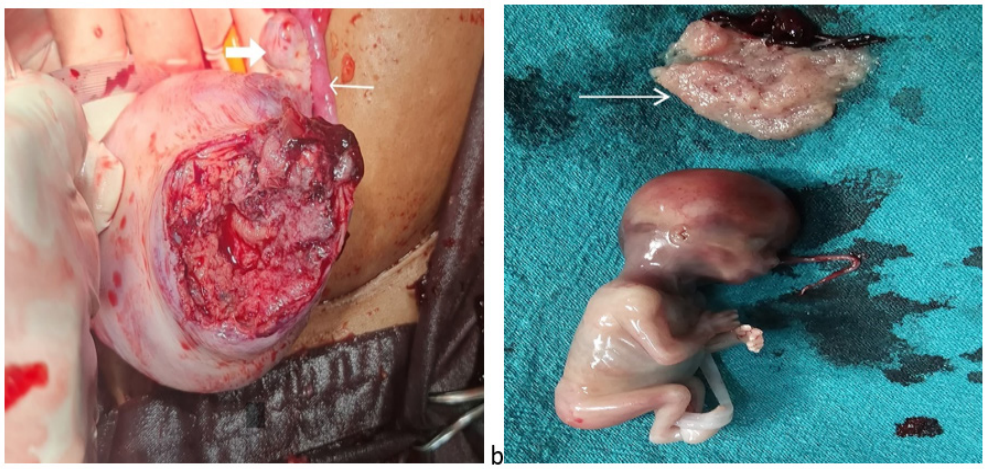

Figure 2: (a) huge defect of left uterine horn above the utero-tubal junction, thick arrow shows the left ovary and the thin arrow showed fallopian tube, (b) the fetus removed from the abdominal cavity and part of placenta removed manually from the ruptured uterine wall indicated by arrow.

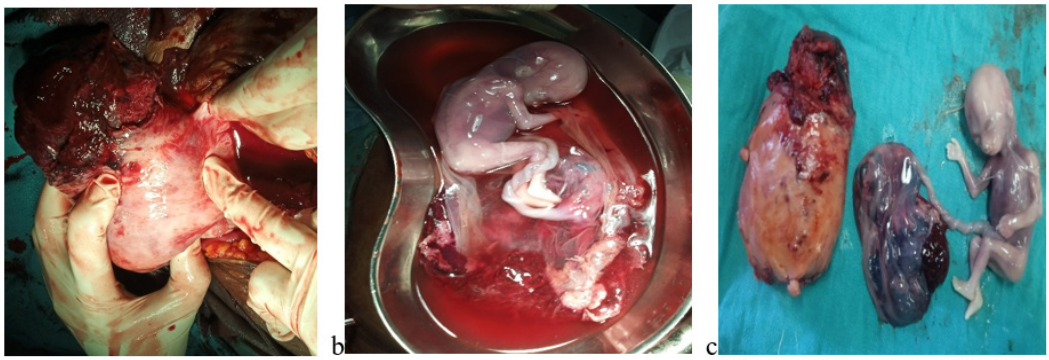

Figure 3: (a) Ruptured right horn of uterus, (b and c) indicates intact gestational sack and fetal extracted from abdominal cavity.

Discussion

Interstitial ectopic pregnancy is an unusual type of ectopic pregnancy, but has 6–7 times higher mortality rate than other ectopic pregnancies [4,5,8]. The increasing number of risk factors such as artificial reproductive techniques, previous tubal surgeries leads to increased incidence [13]. It's implantation in a highly vascularized area and strong distensible submucosal tissue lead to rupture at a later gestational age causing severe torrential hemorrhage [5,8,13]. The rupture rate of interstitial ectopic pregnancies was approximately 15% and the majority of rupture occurs before 12 weeks of gestation [4,5]. The gestational age of the interstitial ectopic pregnancy is related to the degree of hemodynamic instability because of the larger torn of uterine wall was involved with bigger gestation. In our cases, the rupture occurred at 16 weeks gestation and the bleeding was so torrential that threatens the life of the patients.

A ruptured interstitial ectopic pregnancy does not have uniform symptoms and can mimic other conditions and severe complications may develop due to delay in diagnosis [2,4]. Patients may have complaints of dizziness, fainting, abdominal pain, shoulder pain, diarrhea, vomiting, rectal pressure or defecation pain, amenorrhea, vaginal bleeding and urinary symptoms [4,14]. Similarly, our first case presents a history of three episodes profuse watery diarrhea, vomiting, severe abdominal pain, and fainting of less than 24 hours, while the second case started to have vaginal bleeding followed by diffuse abdominal pain of three days. On physical examination, the patient may be in hypovolemic shock with signs of peritonitis and tenderness of cervical motion due to peritoneal irritation. Similarly, our cases present in hypovolemic shock with cold extremities feeble peripheral pulse, un-recordable blood pressure and signs of peritonitis. Interstitial ectopic pregnancies pose a unique diagnostic challenge due to their apparent location [4]. Ultrasound criteria used to diagnose inter-

stitial ectopic pregnancies in the first trimester have a detection sensitivity and specificity of 80% and 98%, respectively [4]. When there is uncertainty, 3D ultrasound and magnetic resonance imaging can be used to confirm the diagnosis [4,15]. In our patients there was massive hemoperitonium, disfigured uterine funds, and non-lived fetus located in the abdomen in both cases. Diagnosis of ruptured uterus was made, resuscitation with crystalloids and blood products was initiated immediately, and patients were prepared for immediate laparotomy, deferring other imaging techniques like MRI for confirmation.

The management of interstitial ectopic pregnancy is based on the clinical condition of the mother and future fertility problems [2,3]. Traditional treatment of interstitial ectopic pregnancy with hemoperitoneum is laparotomy with cornual resection or hysterectomy, in a severely damaged uterus [1,3,13]. Laparotomy or laparoscopy may be used for conservative surgical management. In our cases, a midline incision was made to facilitate fast entry and adequate exposure. In the first case, the defect on the left side of the fundus was thoroughly assessed after cutting the edges containing conceptus products. Repair was performed in three layers with delayed absorbable. Hysterectomy was considered, but with limited blood on hand the procedure was delayed to avoid extra blood.

Although in the second case the condition of the mother was serious with hemodynamic instability, the rupture was extensive, the ongoing bleeding was not easily controllable and completed family which favors subtotal abdominal hysterectomy. Direct injection of diluted vasopressin into the highly vascularized area during conservative surgical measures such as cornual resection may to reduce blood loss but in our case repair alone secures hemostasis [13]. Although rarely used, the regimens for medical treatment of interstitial pregnancy remain unknown, but local injection or systemic therapy with methotrexate, local injection of potassium chloride indicated in hemodynamically stable, early gestation, and diameter less than 4 mm, serum ß-hCG levels less than 10.000 IU/L, and those who can adhere follow up strictly follow-up with serum beta hCG [9,13,15]. Some studies have shown that local, systemic or both, is effective with an overall success rate of more than 83% [1,8].

Follow-up is an important part of treatment, and both medically and surgically treated patients should include serial B-HCG monitoring to exclude the persistence of trophoblastic tissue, which if not treated can progress to gestational trophoblastic disease. In our cases, the edges and bed of the defect with possible trophoblastic tissue were removed in the first case and both patients were followed with serial B HCG which becomes negative in one month.

Conclusion

To avoid morbidity and mortality related to interstitial pregnancy, a high index of suspicion and prompt action are necessary. Diagnosis presents challenges, as they can present with pure gastrointestinal symptoms with or without vaginal bleeding. Bedside ultrasound in ruptured cases of interstitial ectopic pregnancy is sufficient to make a decision.

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