A Case of Ruptured Full Term Interstitial Pregnancy With A Live Mother And Baby*

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ABSTRACT

Interstitial pregnancy is a form of ectopic pregnancy in an unusual location, implanting on the intramural part of the fallopian tube. Because the myometrium is highly distensible, it may allow an interstitial pregnancy to advance up to 16 weeks where it usually presents with rupture. Its late diagnosis and severe hemorrhagic complication accounts for a higher mortality rate compared to other ectopics. On the other hand, interstitial pregnancies that progress to term or near term are extremely rare. From the 10 cases published in literature reporting the delivery of a live term or near term fetus, only 1 of these cases has antenatally diagnosed the presence of interstitial pregnancy prior to rupture by investigating a probable placenta accreta found on ultrasound. This report discusses a case of a ruptured full term interstitial pregnancy diagnosed intraoperatively which resulted to a live mother and baby, and describes retrospectively the similar ultrasound findings of placenta accreta which was realized after rupture.

Keywords: live full term, ectopic pregnancy, interstitial pregnancy,cornual pregnancy, ultrasound, placentaaccreta

INTRODUCTION

Ectopic pregnancy accounts for approximately 2% of all pregnancies and is the most common cause of pregnancy related mortality in the first trimester.[1] The risk of maternal death is 10 times that of childbirth and 50 times greater than first trimester abortion.[2] It occurs when a blastocyst abnormally implants outside the endometrium of the uterus and the majority of which adheres to the different parts of the fallopian tube. Other types, termed as "ectopic pregnancy in unusual locations" include interstitial, cornual, ovarian, cervical, scar, abdominal and heterotopic pregnancies.[1] (Figure 1)

Interstitial and cornual pregnancy accounts for 1-3% of ectopic pregnancies.[3] The interstitial part of the fallopian tube is the proximal portion which lies within the muscular wall of the uterus near its cornu.[4] Some authors use the term cornual and interstitial pregnancies to be synonymous, but others reserved the term cornual pregnancy for gestations in one horn of a bicornuate or a septate uterus.[5] (Figure 2) While the overall mortality rates for ectopic pregnancies over the past five decades have steadily decreased to 0.14%, the case mortality for interstitial pregnancies remains the same which is nearly 15 times higher than other ectopics and still accounts to about 30% of all maternal deaths due to ectopic pregnancies despite its rarity.[6]

Delayed diagnosis of interstitial pregnancy is the main factor contributing to high maternal mortality rate in comparison to other tubal ectopics.[7] The intramural

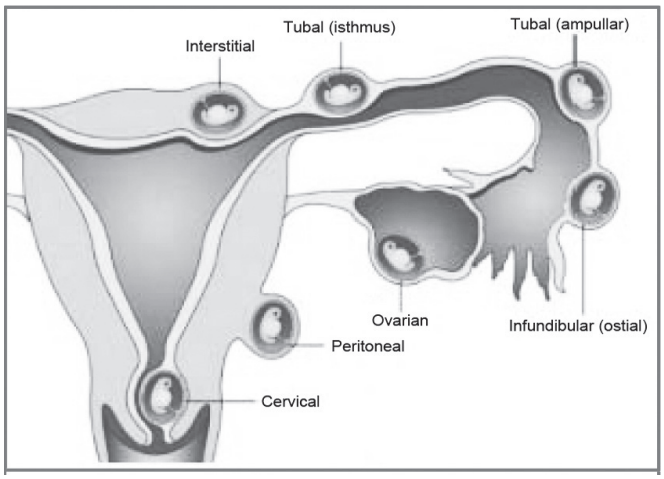

Figure 1. Types of ectopic pregnancy[26]

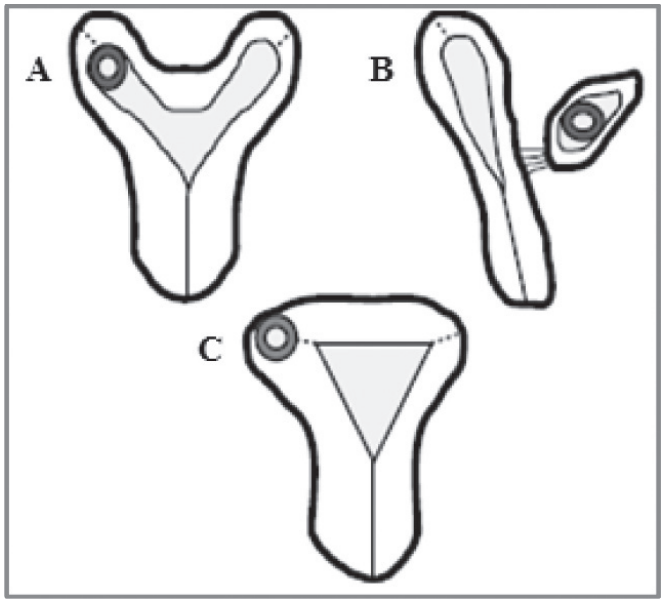

Figure 2. Cornual (A & B) vs.Interstitial pregnancy[7]

* Finalist, 2013 Philippine Obstetrical and Gynecological Society (POGS) Interesting Case Paper Contest, September 19, 2013, 3rd Floor, POGS Building, Quezon City

portion of the fallopian tube allows a substantial degree of distensibility, hence, in contrast to a tubal ectopic pregnancy which usually ruptures at 6-8 weeks age of gestation,[8] an interstitial pregnancy may progress without symptoms until 7-16 weeks where catastrophic complications such as uterine rupture occur.[4] Because it is located in the most richly vascularized site of the female pelvis, within the junction of the branches of the uterine and ovarian arteries, hemorrhage may be massive and life threatening, noting to be 2.5-5 times greater than other ectopic pregnancy locations. Rupture may immediately lead to hypovolemic shock and death.[9,4] Early diagnosis and treatment, therefore, are essential in reducing maternal mortality.[4] Identification of risk factors, a high degree of clinical suspicion for any untoward signs and symptoms and familiarization with diagnostic clues on ultrasound will aid for its timely detection and treatment.

Interstitial pregnancy is usually diagnosed once rupture occurs or detected by certain early sonologic findings. Once caught, the definitive treatment is termination of pregnancy which may be done surgically or medically. Expectant management is not favored, because viable pregnancies are extremely rare and complications are life threatening. Although, in the 10 reported occurrences in literature according to the research of Scarella, et. al, an interstitial pregnancy advanced to term or near term and brought about a live fetus. There were 4 cases of full term live births and 6 preterm where 2 of which died of respiratory distress. Only 1 of these reports has successfully antenatally diagnosed an interstitial pregnancy prior to rupture by incidentally finding a possible placenta accreta on ultrasound and was further investigated using MRI.[4] In this paper, we report another rare case of a full term interstitial pregnancy which resulted in a live mother and baby which was diagnosed intraoperatively. We also describe in retrospect similar ultrasound findings of placenta accreta which was only realized after rupture.

GENERAL OBJECTIVE

To present an unusual case of a full term interstitial pregnancy with a live mother and baby

SPECIFIC OBJECTIVES

1. To discuss a case of a full term pregnancy presenting as acute abdomen and incidentally diag nosed as interstitial pregnancy intraoperatively
2. To discuss about the epidemiology, risk factors, diagnosis and treatment of interstitial pregnancy
3. To compare the ultrasound findings of the only successful case of antenatally diagnosed advanced interstitial pregnancy with that of the patient

CASE HISTORY

This is a case of R.G., a 28 year old gravida 2 para 1 with an obstetric score of 1-0-0-1, single with a live in partner, Filipino, Roman Catholic, who was admitted last March 21, 2013 at 11:45 pm with the chief complaint of abdominal pain.

Patient's past medical and family histories were both unremarkable. She is a non-smoker, non-alcoholic beverage drinker, a high school graduate and a housewife. Menses started at 11 years old with irregular cycles, lasting 5-7 days, using 4-6 pads per day without associated signs and symptoms. Her last menstrual period cannot be recalled. Sexual debut started at 17 years old with 2 sexual partners. The first one lasted for 2 years, an electrician, who was apparently monogamous and the second, her current partner, an overseas worker, whom she has been with for 6 years, is also apparently monogamous. During their first few months together, the patient used oral contraceptive pills but discontinued when they decided to live together. They were trying to conceive for 2 years but was unsuccessful. There was a history of passage of yellowish foul-smelling vaginal discharge 2 years prior to admission which was relieved by unrecalled antibiotics prescribed by a doctor. The patient then had spontaneous conception of her eldest child who is 1 year and 7 months old who was delivered full term via normal spontaneous delivery in our institution without complications.

Prenatal history revealed 2 incidences of hospital admissions in different institutions during the present pregnancy. Both were due to abdominal pain which were diagnosed as urinary tract infection with threatened preterm labor and treated as such by unrecalled medications. The first confinement was 5 months prior to admission where she experienced moderate hypogastric pain without vaginal bleeding. A pelvic ultrasound was done showing a single, live, intrauterine pregnancy at 13 4/7 weeks age of gestation with normal amniotic fluid and circumferential placenta without retroplacental hematoma. Patient was then discharged improved and shesubsequently continued her prenatal check-up at their local health center. Then, the second confinement happened 2 months prior to admission, when the abdominal pain recurred. A repeat pelvic ultrasound was performed showing a single, live, intrauterine pregnancy at 34 5/7 weeks age of gestation, in breech presentation, with a posterior low lying placenta (Figure 3) and a male fetus weighing 2400 grams. Patient was again discharged and was advised to continue her prenatal check up with an obstetrician for the possibility of a cesarean delivery due to malpresentation and a low lying placenta but was lost to follow up; until 1 day prior to admission, the patient decided to consult our institution's out-patient department where she was requested

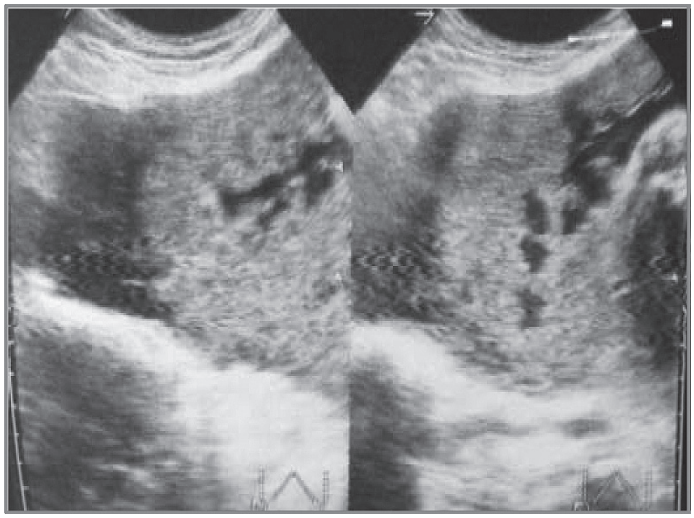

Figure 3. Ultrasound at 34 weeks AOG

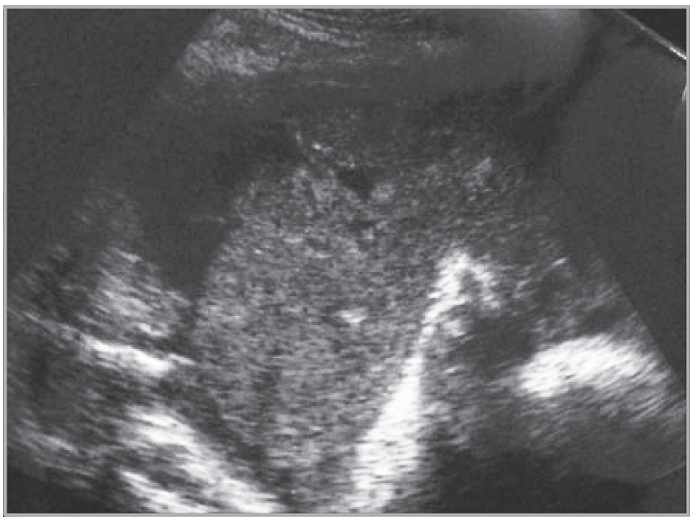

Figure 4. Ultrasound on admission

a repeat ultrasound which revealed a single, live, intrauterine pregnancy, 37 3/7 weeks age of gestation by biometry, full term by non biometric parameters, 2800 grams in frank breech presentation, with a high lying placenta, and adequate amniotic fluid. The patient was then advised regarding trial of labor and encouraged regular follow up.

The history of present illness then started about 2 hours prior to consult when the patient experienced sudden moderate abdominal pain while watching television. It was located all over the abdomen, continuous, increasing in severity, and aggravated by movement. There was no associated vaginal bleeding or discharge and there was good fetal movement. The persistence and progressively increasing severity of the pain prompted consult in our institution's emergency department. The patient arrived wheelchair-borne, sweating all over and was crunching on her abdomen complaining of severe abdominal pain. Vital signs were as follows: blood pressure of 110/80 mmHg, heart rate of 77bpm, respiratory rate of 28cpm, and temperature of 37. She was not pale-looking, and had pink palpebral conjunctivae. Respiratory and cardiovascular systems were unremarkable. The abdomen was globularly enlarged with normal fetal heart tone. It was noted to be rigid on palpation, with moderate direct & rebound tenderness all over. Estimated fetal weight by mapping was 2600-2800 grams. On internal examination, the cervix was closed and uneffaced with no cervical motion tenderness. There was no vaginal bleeding or discharge. The impression then was: G2P1 (1001) Pregnancy, uterine, 37 4/7 weeks age of gestation by ultrasound, breech, not in labor, Acute abdomen probably ruptured viscus versus abruptio placenta.

Intravaneous access was then initiated. Blood was extracted and sent for laboratory examination. Hemoglobin was 10.2 g/dL, with normal hematocrit, leukocytes, and platelet count. Ultrasound was done revealing normal fetal heart tone but there was note of a high lying placenta with areas of lucency and a large hypoechoic area immediately superior to it (Figure 4). The patient was then sent to the operating room for an emergency exploratory laparotomy and primary low segment cesarean section. Under spinal anesthesia, a midline infraumbilical incision was made. Hemoperitoneum of 500 cc was obtained. The lower uterine segment was unformed while the middle aspect was thinned out. A vertical uterine incision was then made and a live baby boy was extracted in breech presentation with an APGAR score of 8, 9, pediatric aging of 37 weeks, weighing 2400 grams which is appropriate for gestational age (Figure 5). Afterwhich, a very thin sac-like structure was left where the placenta was adherent and this was continuous with the thick lower part of the uterus (Figure 6). Its posterior aspect containeda 1 cm perforation (Figure 7) while there were also numerous engorged blood vessels which bled profusely after clamping of the placenta yielding a blood loss of 2 liters (Figure 8). The fallopian tubes and ovaries were grossly normal (Figure 9). There were no adhesions noted. Hence, with only 1 unit of packed RBC available for transfusion, total hysterectomy was done.

On inspection of the specimen, the sac with the placenta contained large blood vessels and is adherent to the left side of the uterine fundus (Figure 10 & 11). On its posterior aspect, the myometrium of the fundus seemed to have stretched and fused with the membranes (Figure 12). On cut section, the endometrial cavity is filled bydecidual tissue. The interstitial part of the left fallopian tube is seen communicating with the sac (Figure 13). With these, the diagnosis of a left interstitial pregnancy with placenta increta was attained. Subsequent histopathologic examination confirmed this diagnosis.

Estimated blood loss with the procedure was 3 liters.

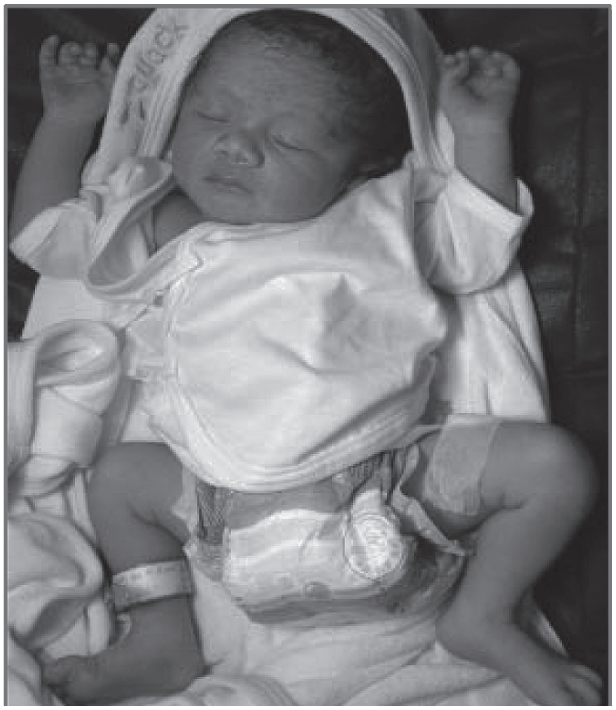

Figure 5. Live full term baby

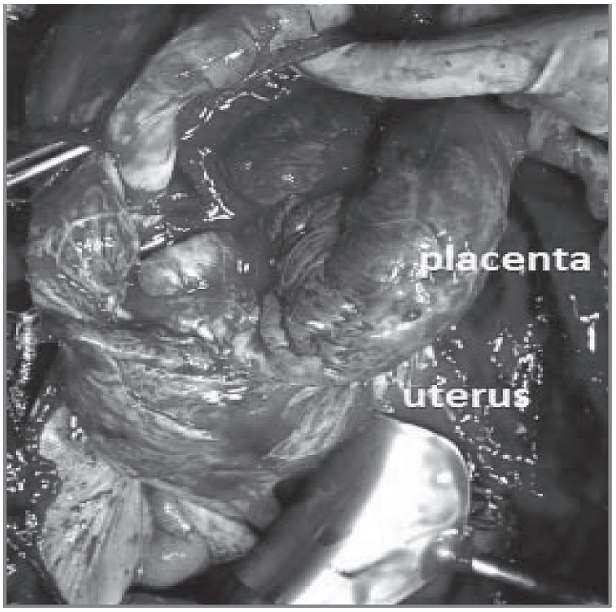

Figure 6. Placenta adherent to the uterus

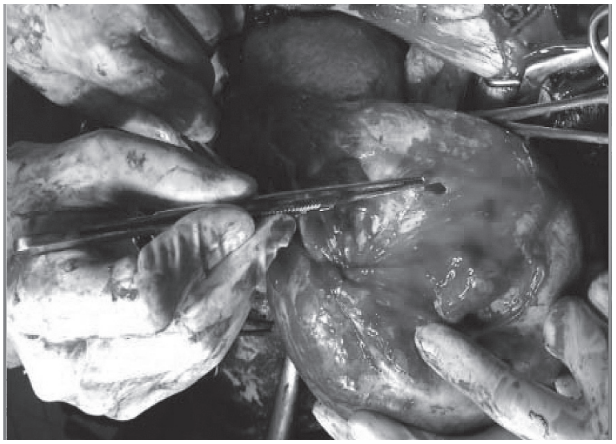

Figure 7. Sac with perforation

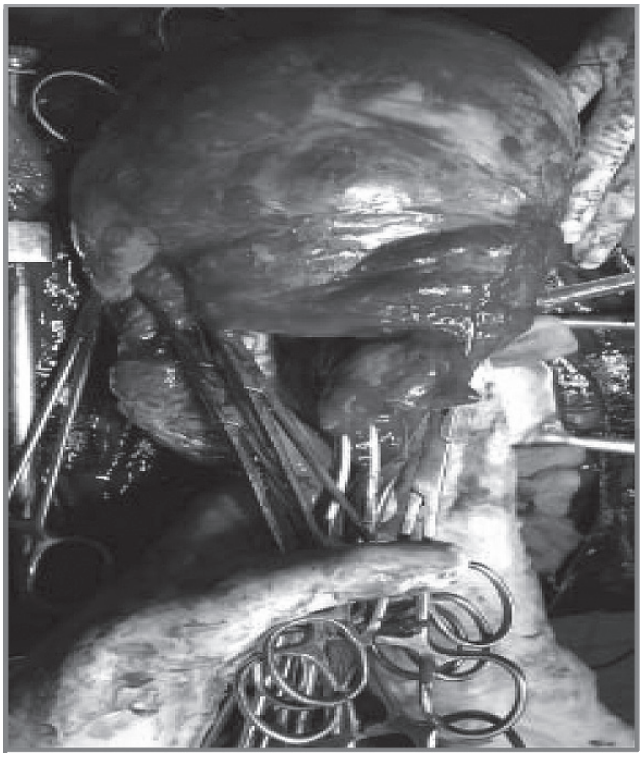

Figure 8. Bleeding vessels

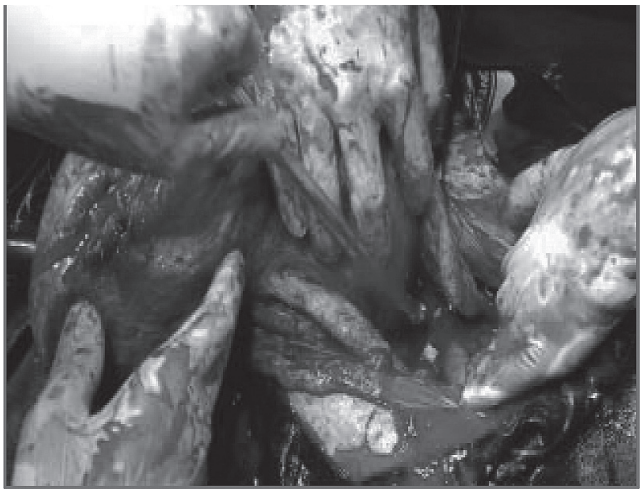

Figure 9. Ovaries and fallopian tubes

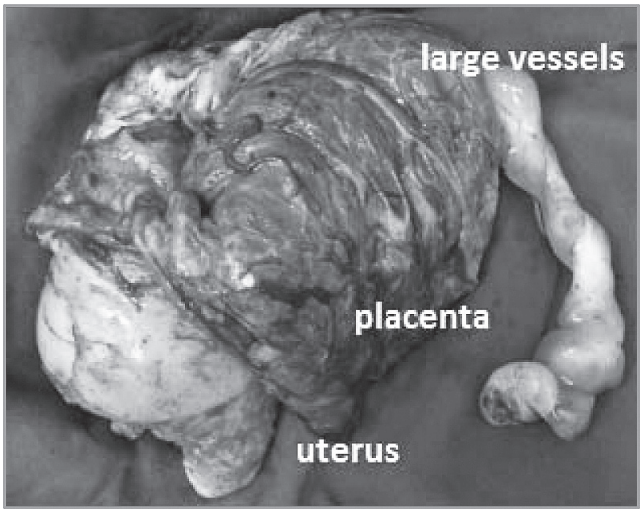

Figure 10. Dilated vessels

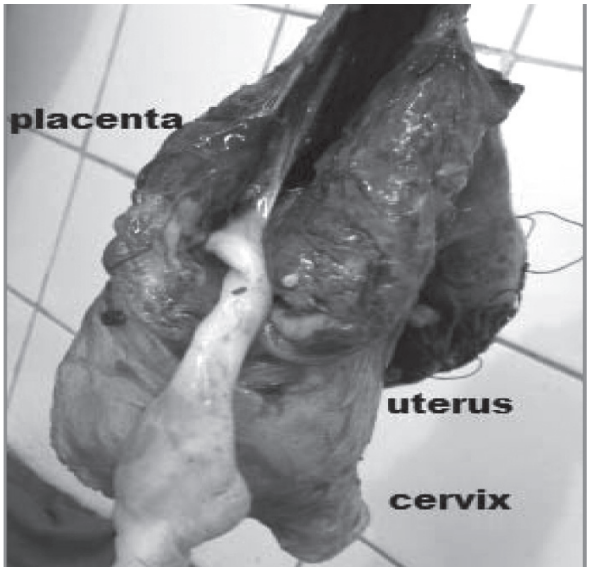

Figure 11. Placenta attached to left fundal area

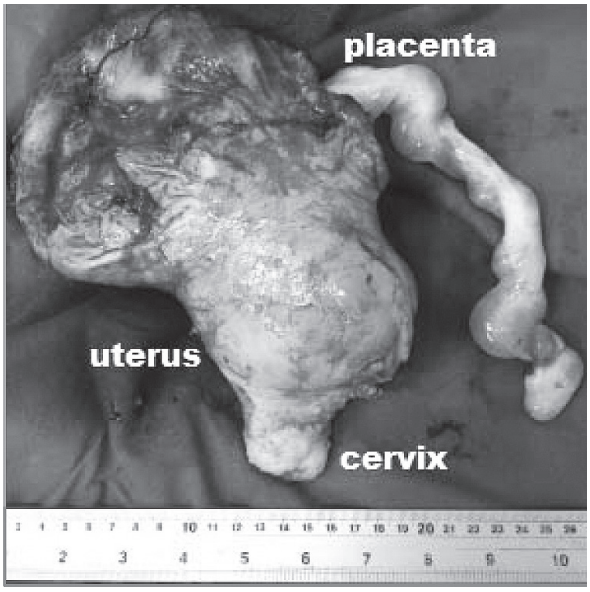

Figure 12. Myometrium stretched up

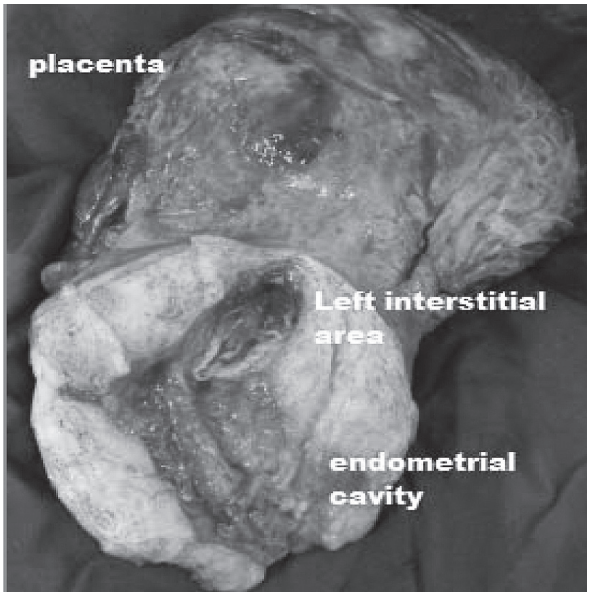

Figure 13. Endometrial cavity & left interstitial area

Patient was transfused with 1 unit packed RBC and given 2 liters of crystalloid solution and 1 liter of colloids. Blood pressure during the OR was 100-80/60-40 mmHg and heart rate ranged from 90-110 bpm. Urine output was adequate. Patient was started on antibiotics and pain relievers. Afterwards, patient was sent to the post anesthesia care unit where she was noted to be conscious and coherent and had stable vital signs. Patient was transfused with 1 more unit packed RBC. A repeat hemoglobin was requested yielding a value of 9g/dL. Patient was then sent to the ward's intensive monitoring unit.

Twelve hours post operatively, patient was started on general liquid diet, venoclysis was continued, and IV medications were shifted to oral. The wound was dry, and there was scanty vaginal bleeding. Vital signs remained normal. On the first hospital day,a repeat hemoglobin was requested revealing a value of 7.6 g/dL. Hence, transfusion of 3 more units packed RBC and 2 units fresh frozen plasma was requested. Due to the inavailability of blood products, the patient was only transfused with 2 more units of pRBC until the 5th hospital day, and was discharged improved on the 6th hospital day with a hemoglobin of 9.8 g/dL and given home medications.

CASE DISCUSSION

We are presented by a case of a 28 year old gravid 2 para 1 (1-0-0-1) who has a history of primarily infertility and pelvic infection, both of which are known risk factors for ectopic pregnancy. Inflammation of the fallopian tubes or salpingitis, is the encountered cause in 50% of ectopics, increasing its risk sevenfold.[2] Previous salpingitis causes permanent agglutination of the plicae of the endosalpinx which may prevent or delay the normal transport of a morula in its 7-day passage to the fallopian tube. Any lag in its transit may lead to the wrong implantation of the blastocyst causing ectopic pregnancy such as previous tubal surgery, use of intrauterine device, conception with assisted reproductive techniques and intake of oral contraceptive pills. Commonly, the blastocyst burrows through the epithelium of the distal fallopian tube and implants within its muscular wall. As the rapidly proliferating trophoblast erodes the subjacent muscularis layer, blood vessels are disrupted. Externally, vaginal spotting may happen and within the fallopian tube hematoma forms which causes the stretching of the peritoneum causing episodic pain before the final perforation into the peritoneal cavity. Because of limited space and inadequate blood supply, rupture occurs as the serosa is maximally stretched, producing necrosis.[10] In our case, the implantation within the interstitial portion of the fallopian tube offered an abundant muscle layer within the uterine fundus providing a potential space for growth and a vast blood supply from

the ovarian and uterine arteries to adequately nourish an enlarging fetus. In relation to an advanced abdominal pregnancy, which is similarly a rare and interesting phenomenon encountered in 1 in 25,000 births[11], the abdomen may provide a huge potential space for fetal growth but the absence of a definite vessel to contain it may easily cause compression and commonly produce deformities, as opposed to interstitial pregnancies where the myometrium likewise serves as protection from external pressure. Also, the distensibility of the myometrium may not allow the classic symptoms of ectopic pregnancy to beas prominent. In our patient, the presenting symptom was recurrent abdominal pain without vaginal bleeding which may have been significant enough to prompt hospital admissions, but its resolution and prompt response to treatment did not allow the underlying pathology to be investigated further.[4] Hence, the subtleness of the signs and the rare occurrence of interstitial pregnancy, makes clinical diagnosis difficult.[6]

Sonographic diagnosis, on the other hand, is also equally challenging. Since this type of ectopic pregnancy is very rare, perhaps only a few sonographers have encountered such cases during their lifetime.[7] As in the case of our patient, the diagnosis was missed on her previous hospital admissions. Because of this problem, some authors have suggested sonologic clues compiled from previous literature to help in its diagnosis, but unfortunately, most of them apply only to early interstitial pregnancies. Since only 10 cases of term and near term interstitial pregnancies have been reported in literature, no sufficient data can be gathered to accurately diagnose this.[4] Current knowledge surrounding these remains largely observational and anecdotal.[3] Scarella et. al. has reported the first published case of a successfully antenatally diagnosed advanced interstitial pregnancy at 20 weeks age of gestation (as of March 2012). They were able to detect the presence of placenta accreta seen as placental lacunae with partial loss of clear space between the myometrium and placenta (Figure 14). Hence an MRI was done which showed an interstitial pregnancy. Although, they opted to terminate the pregnancy as soon as it reached 28 weeks in order to avoid rupture but this resulted in fetal demise from respiratory distress 12 hours after.[4] The same sonologic and histologic findings were seen in our case. The sonoluscent areas seen within the placenta which was overlooked prenatally represent vascular lakes which are much commonly seen in placenta accreta/increta in a scarred uterus complicated by placenta previa. Enlargement of the blood vessels happens when the placenta adheres to a site without an intervening deciduabasalis. They are described as dark areas with swiss cheese or moth-eaten appearance on ultrasound (Figure 15). A color Dopplerultrasound or

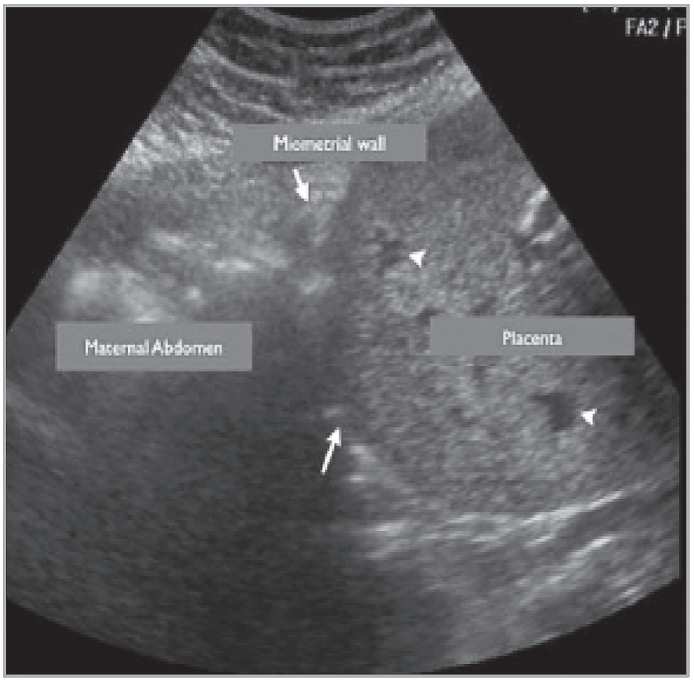

Figure 14. Ultrasound findings of accreta by Scarellaet. al.[4]

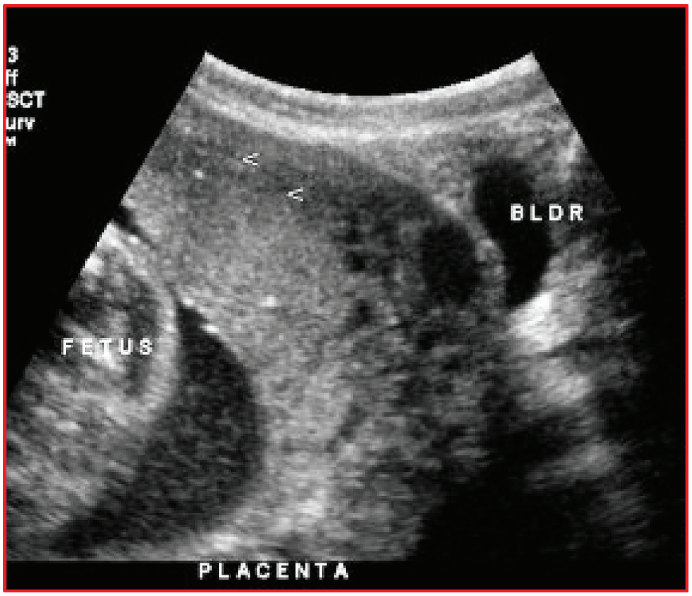

Figure 15. Usual ultrasound findings in placenta accreta with placenta previa[12]

MRI isusually suggested to confirm its presence.[12] Since placenta accreta can also predispose to severe hemorrhage, its prenatal diagnosis would enable the obstetrician to prepare. Hence if placenta accreta was considered in our patient prior to rupture, MRI would have diagnosed the presence of interstitial pregnancy and blood products could have been secured to anticipate serious blood loss. And in the advent of sudden rupture, availability of blood products could have avoided a hysterectomy.

The management of interstitial pregnancydepends on many factors including the desire for future pregnan-

cies,[13] gestational age, hemodynamic status and presence of uterine rupture.[14] If the interstitial pregnancy is correctly identified at an early stage, medical management or laparoscopic resection can be offered.[8] On the other hand, for much more advanced interstitial pregnancies and for those with symptoms of hemorrhage, laparotomy is recommended in the form of cornual resection or hysterectomy. Resection of the cornual region would produce a full thickness uterine incision and is associated with decreased fertility rates and also with uterine rupture in up to 32% of cases.[11, 9, 15] This procedure adds up to the already diminished chance of fertility seen in patients with previous ectopics, with up to 28% probability of recurrence and only one third chance of delivery of a liveborn infant.[16] Hence, even though an interstitial pregnancy was not diagnosed in our patient before rupture, the birth of a healthy second child in exchange for her fertility may have spared her from the possibility of recurrence of another ectopic pregnancy. Or even if she bears another pregnancy, the probability of uterine rupture after cornual resection is high which can be equally dangerous.

On the other hand, sincerupture has occurred producing life threatening hemorrhage, hysterectomy ultimately becomes the only treatment.[14] Although the distensibility of the myometrium has eventually reached its limit leading to rupture, in our case, it has allowed the fetus to grow to term and culminated in a minute perforation, producing abdominal pain severe enough to initiate a timely consult and detect the need for immediate surgical intervention but was also small enough to delay massive hemorrhage. Hence, the tear in the uterus, happening after the time of almost certain fetal survival, a tear small enough to point out that a pathology was present, the tear which signalled the appropriate timing of delivery of an infant that cannot be born naturally, may have just been the tear which has saved the life of the mother of the fetus. This tear that was thought to only bring about death turned out to be the tear that has become the source of joy.

CONCLUSION

With the increasing number of sexually transmitted infection and assisted reproductive techniques, the incidence of ectopic pregnancies in unusual locations is bound to increase[15]. Since only limited data are available, case reports and anecdotal evidence such as what was described in this case could potentially be of great benefit in formulating new diagnostic approaches which could hopefully help prevent catastrophic complications including maternal and fetal mortality and preserving fertility.

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