

Undiagnosed asymptomatic second trimester broad ligament ectopic pregnancy: a case report and mini-review

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Abstract

Pregnancy in the broad ligament is a retro-peritoneal abdominal pregnancy. It is a rare form in which the gestational tissue gets implanted between the layers of the broad ligament. Due to its rarity, the diagnosis is infrequently established before surgical intervention. A twenty-five year old nulliparous woman presented for routine antenatal care in the second trimester (± 17 weeks). Routine ultrasound revealed an empty uterus with a large pelviabdominal mass. This picture suggested and undiagnosed extra-uterine pregnancy. A right broad ligament ectopic pregnancy was diagnosed at exploratory laparotomy, excision of pregnancy and right salpingectomy was performed. Postoperative course was uneventful and serum hCG was undetectable at the fourth week after surgery. Information about this rare form of ectopic pregnancy is observational and mainly comes from reported cases. A high index of suspicion is needed for early diagnosis and intervention.

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Introduction

Ectopic pregnancy (EP) is one of the major causes of maternal mortality, with a reported incidence reaching 10-15% of all maternal deaths.¹ Abdominal EP is a rare highly morbid form of ectopic pregnancy in which implantation may occur in the omentum, vital organs or large blood vessels. Broad ligament EP is a subtype of abdominal pregnancy entailing implantation of the gestational tissue between the leaves of the broad ligament.^{2,3}

There is a great variation in the clinical presentation of this rare form of EP; ranging from being asymptomatic to rupture in labor at term. Absence of reliable clinical features makes the diagnosis more challenging and requires a high index of suspicion from the clinicians. In most cases, the diagnosis is delayed up to advanced gestational age,⁴ and is rarely established before surgery. Once the diagnosis of broad ligament EP is suspected, surgery

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should be performed.^{2,5}

We report a case of spontaneous broad ligament EP in a completely asymptomatic woman who presented in the second trimester for routine antenatal care. A review of reported cases of broad ligament EP extended to the second trimester is included in the discussion.

Case Presentation

A 25 year-old G2P0+1 pregnant woman at \pm 17 weeks gestation (by sure dates) was referred to Assiut Women's Health Hospital, Upper Egypt for ultrasound evaluation of her current pregnancy. She gave a history of primary infertility of two years duration which was followed by a spontaneous right EP (tubal) one year ago. She subsequently received two doses of systemic methotrexate which led to complete resolution of her EP.

She presented to the ultrasound (US) unit with an ultrasound report (performed one week before in her village) indicating that: she had a single living fetus with an average biometry of 16 weeks, marked reduction of amniotic fluid and a huge placenta compressing the fetal parts. Clinical examination showed normal vitals, BMI 23 Kg/m², and no abdominal tenderness. She had no history of vaginal bleeding but there was a history of previous treatment of threatened abortion at 9 week's gestation. Per vaginal examination revealed a closed cervical os and a pelvic-abdominal mass of about \pm 14

week's gestation in size; which was assumed to be the gravid uterus. No definite tenderness was elicited.

Surprisingly, detailed abdominal and transvaginal US examinations revealed an empty normal sized uterus which was pushed anteriorly by a pelviabdominal mass nearly 11x8x6 cm in size with heterogeneous consistency (Figure1). Neither definite fetal parts nor cardiac activity could be seen. Minimal pelvic collection was detected. An impression of undiagnosed extra-uterine pregnancy was strongly suspected. Laparotomy through Pfannenstiel incision was done under general anesthesia after counseling about her current condition and taking an informed consent.

During exploratory laparotomy, no hemoperitoneum was found; however, a right large, bluish broad ligamentary mass was seen (~12 cm) with few omental adhesions and a markedly stretched right tube and round ligament. There was no sign of rupture. The right broad ligament was opened and this revealed a dead fetus with crowded parts, placenta, blood clots, and fetal membranes (Figure 2). A diagnosis of right broad ligament EP was established. The right tube was ruptured and damaged during extraction of firmly adherent placental tissues; hence, right salpingectomy was carried out. Continuous oozing from the right broad ligament necessitated ligation of the right uterine vessels to achieve hemostasis. Estimated blood loss was~700 ml.

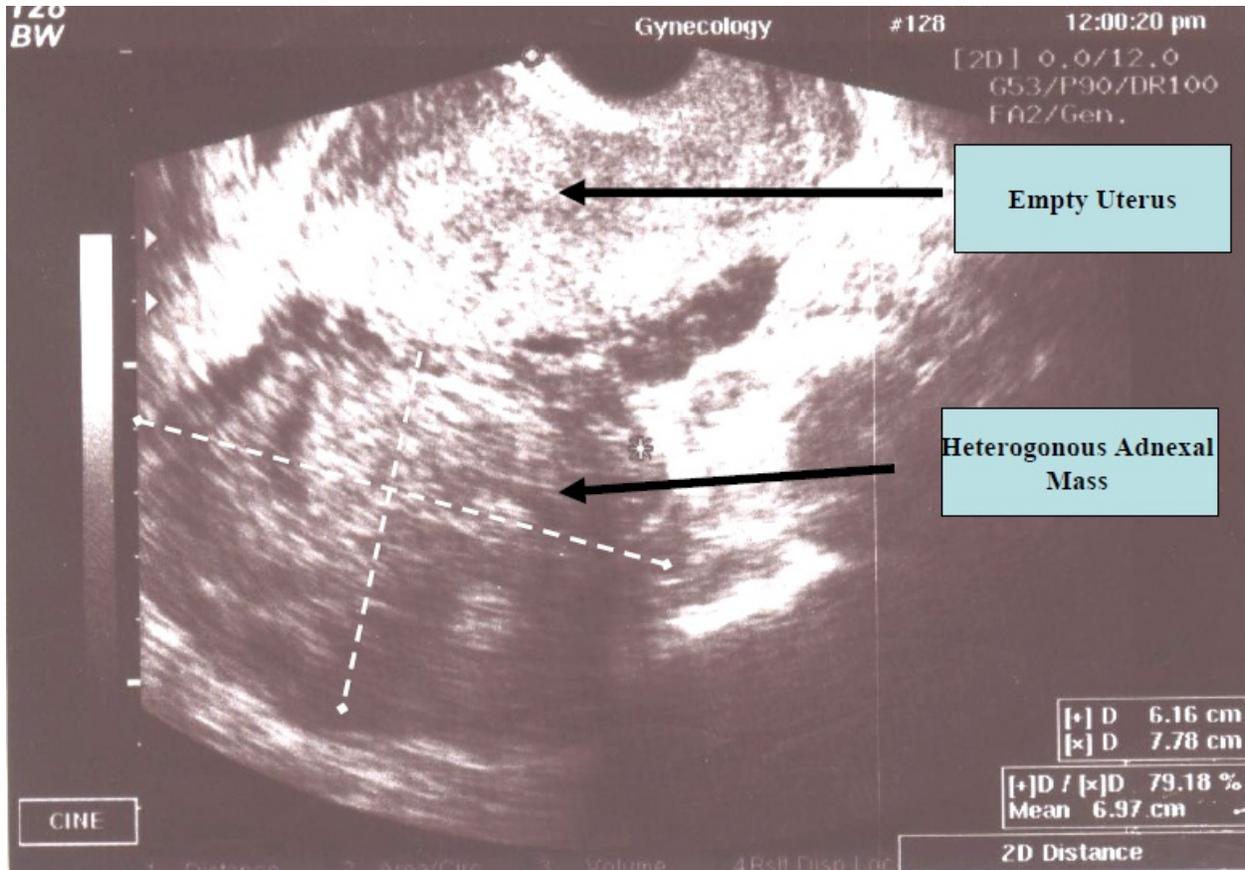


Figure 1: Abdominal ultrasound showing empty uterus with right heterogeneous adnexal mass pushing the uterus anteriorly.

The patient received 2 units of packed RBCs during the operation. She was subsequently transferred to the postoperative care room after urethral Foley's catheter fixation and administration of 2 gm of cephalosporin.

The patient had an uneventful recovery and was discharged on the third postoperative day. Her serum hCG was undetectable on the fourth week of follow-up visits.

Discussion

Broad ligament ectopic pregnancy is a rare type of abdominal EP especially

with spontaneous conception. The reported incidence of all abdominal pregnancies is 1.4% of ectopic pregnancies.⁶ Previous history of pelvic inflammatory disease, EP, pelvic surgery and use of assisted reproductive techniques (ART) in treatment of infertility are considered significant risk factors for development of such EP.² The definite pathogenesis for the development of broad ligament EP is unknown. It is believed that it may result from primary implantation of the fertilized ovum in the broad ligament or secondary implantation from ruptured tubal pregnancy.^{7,8}

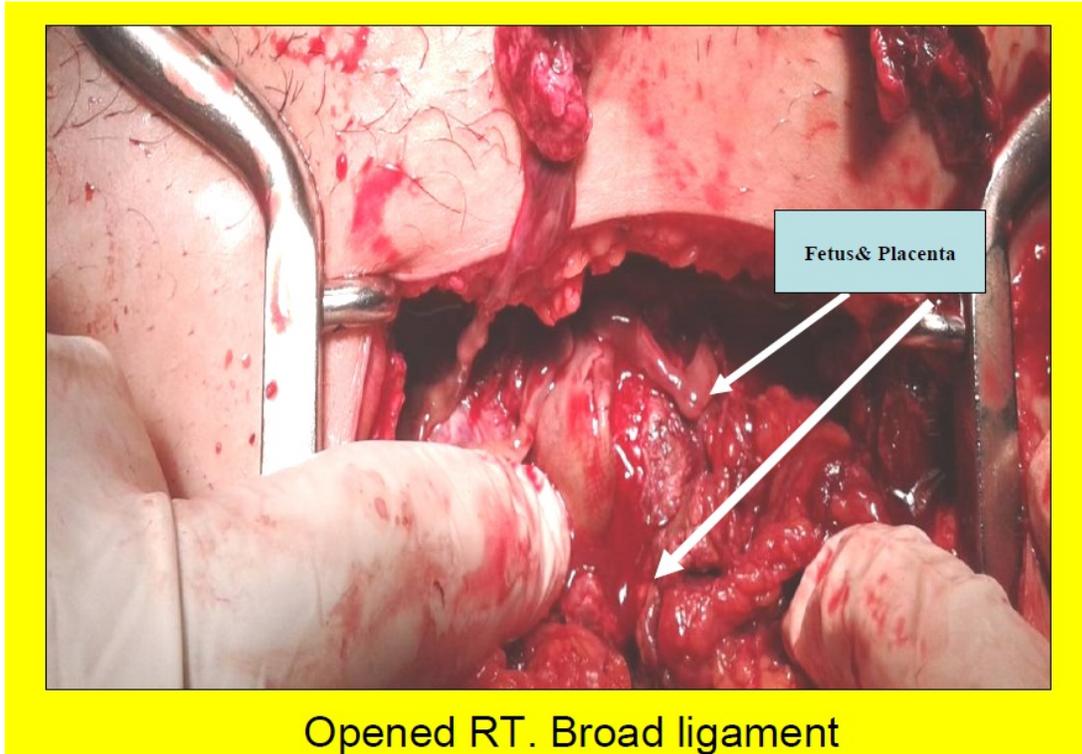


Figure 2: Opened right broad ligament during laparotomy revealed dead fetus with placental tissue inside.

Diagnosis of broad ligament EP is rarely established before laparotomy. Many factors are implicated in the late diagnosis. The relative infrequency of this type makes the study of diagnostic features difficult. Similarly, the great variability in the clinical presentation and the possible absence of physical findings reported with this type, such as abdominal tenderness, thickening of the broad ligament, retracted cervix and bulging of the cul-du-sac,² may also contribute to the delay in diagnosis. Even with the use of ultrasound, it may be misdiagnosed as another pelvic pathology such as fibroid.⁹

The absence of identifiable risk factors is commonly cited as a cause of misdiagnosis.¹ Late diagnosis is associated with a high risk of rupture, disseminated intravascular coagulation (DIC), bowel obstruction and fistula.¹⁰ Diagnostic laparoscopy¹¹ and MR¹⁴ are promising modalities that may be valuable in doubtful cases.

Table 1: Summary of cases of second trimester broad ligament ectopic pregnancy published in the literature.

Author & year	Age	Parity	Risk factor	Gestational Age	Presentation	US Findings	Intervention
Chia et al, 1993 ³	17	G1P0+0	Not known	20 wk	Medical termination of pregnancy	Hysterosalpingography showed right extrauterine pregnancy	Laparotomy: right broad ligament pregnancy
Cordero et al, 1994 ²	26	G2P1+0	?? Obesity	18 wk	Abdominal pain	Extrauterine viable pregnancy to date	Laparotomy; left broad ligament pregnancy, no evidence of prior tubal rupture
Abdul et al, 2008 ¹²	33	G7P6+0	Not known	26 wk	Asymptomatic	Non-viable pregnancy in the right iliac region	Laparotomy; excision and unilateral salpingo-oophorectomy
Sharma et al., 2011 ¹³	29	G2P1+0	Abdominal tuberculosis	18 wk	Abdominal pain	Viable pregnancy in the broad ligament	Laparotomy; excision and unilateral salpingo-oophorectomy
Sagili and Rani, 2013 ¹⁴	27	G3P2+0	Not known	20 wk	Abdominal pain	Viable pregnancy in the broad ligament	Preoperative systemic methotrexate then Laparotomy; and excision of pregnancy
Audifred Salomón et al, 2013 ¹⁵	24	G1P0	Not known	15 wk	Abdominal pain	Viable pregnancy in the right broad ligament	Laparotomy; excision of pregnancy
Naeiji et al., 2015 ¹⁶	42	G5P4+0	Not known	14 wk	Abdominal pain	Viable pregnancy in the left adnexa	Laparotomy; excision of pregnancy & left salpingectomy

Management options of this rare form of EP are based on the reported cases only as there are no prospective trials to suggest optimal treatment plans. Surgical removal of the fetus and placenta is the most accepted management option.² Most of the data concerning this unusual form of EP come from cases reported in the literature. To the best of our knowledge, there are only seven reported cases of advanced broad ligament EP reaching up to the 2nd trimester^{2,3,12-16} (Table 1)

The most feared complication encountered during surgical intervention especially in advanced gestational age is hemorrhage as reported by Rahaman⁴ and Cardosi et al.¹⁷ To avoid maternal morbidity, the decision of whether the placenta should be removed or not should be prudent. Ligation of the placental blood supply should be attempted when possible to reduce maternal complications,^{8,10,18} which may occur when expectant or medical management with methotrexate are used until involution.^{8,17} Laparoscopic management is used only in small broad ligament ectopic pregnancies as first reported by Olsen et al.¹⁹ In our case laparoscopic management was not the option of choice because of the relative large size of the EP. The role of methotrexate is very restricted to certain cases. It could be used only in small (<4 cm) unruptured EP in a hemodynamically stable patient.²⁰

In this case, we want to emphasize that careful clinical history and a high index of suspicion are the keys for early diagnosis and prompt management. We believe that reviewing more recent cases about this rare type of EP will

in addition to our case. By reviewing these reported cases, we found that most cases have no identified risk factor and the most common presenting symptom is recurrent abdominal pain followed by being completely asymptomatic. Late diagnosis in these cases facilitates the growth and erosion of gestational tissue into the broad ligament and thus delays the appearance of symptoms. In our case the diagnosis was suspected only by US scan during routine antenatal care.

provide more reliable clinical insight and improve the limited information about it in order to decrease the rate of missed and late diagnosis, improve the outcome and decrease maternal morbidity.

Conclusion:

Broad ligament ectopic pregnancy is a rare form of EP. The diagnosis requires a high index of suspicion especially with the presence of risk factors. Early diagnosis and proper management are vital in order to decrease maternal morbidity.

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