Spontaneous Rupture of the Uterine Artery in an Otherwise Normal Pregnancy

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ABSTRACT

Spontaneous rupture of a uterine artery in pregnancy is associated with a high rate of mortality. Although uterine artery rupture has been associated with postpartum hemorrhage, it is rarely found during pregnancy. Unfortunately, clinical signs and symptoms are usually vague and nonspecific. We report a case of a 36-year-old woman at 20 weeks gestation presenting with abdominal pain who was found to have a spontaneous uterine artery rupture. To our knowledge, this is the first case report demonstrating imaging findings in a patient with this condition. Our patient underwent successful ligation of the uterine vessel with preservation of both mother and fetus. We will discuss possible etiologies of uterine artery rupture during pregnancy, associated imaging findings, and management options.

CASE REPORT

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A 36-year-old G1P0 (gravida 1, para 0) at 20 weeks gestation presented to the emergency department with a 5 day history of suprapubic abdominal pain and a 3 day history of nonbloody emesis and vaginal spotting. Physical examination revealed a woman in mild discomfort with stable vital signs: pulse of 88 beats per minute, blood pressure of 117/72 mmHg, and respiratory rate of 16 breaths per minute. At initial presentation a complete blood count (CBC), and urinalysis (UA) were ordered along with a transabdominal pelvic obstetrical ultrasound to monitor fetal status. CBC noted a mildly elevated white blood count at 13.16x103/UL [reference range 4.50-11.0x103/UL] and a mildly decreased hemoglobin and hematocrit (HGB and HCT) at 10.2 g/dL [reference range 12.0-15.0 g/dL] and 30.9% [reference range 30.0-47.0%], respectively. Urine analysis noted trace blood, leukocyte esterase, and bacteria. No fetal abnormalities were observed on ultrasound (Fig 1). The patient was discharged with a

ay The patient returned to the emergency department 17 hours following discharge, with a reported syncopal episode,

Macrobid and Tylenol.

nausea, and abdominal pain. Repeat physical examination revealed increased heart rate of 95 beats per minute, decreased blood pressure at 108/66 mmHg, and increased respiratory rate of 20 breaths per minute. Repeat CBC at this time demonstrated further elevated WBC count 16.17x103/UL and decreased Hgb 9.7g/dl and HCT 28.9%. A repeat abdominal ultrasound demonstrated anechoic free fluid in the upper quadrants bilaterally with complex echogenic fluid in the pelvis, consistent with hemorrhagic fluid (Fig 2). Obstetrics and Gynecology (OB/GYN) was consulted at this time and an additional follow-up Hgb/Hct was 8.7g/dl and 25.7%. Internal hemorrhage was suspected due to the presence of intraperitoneal fluid and the patient's decreasing hemoglobin and hematocrit. The patient was consented for emergent

diagnosis of a urinary tract infection and sent home with

computed tomography (CT) of the abdomen and pelvis. CT findings demonstrated a 1.4 x 1.3 centimeter (cm) area of contrast extravasation in the left adnexa, suspected to be arising from the left uterine or ovarian artery (Fig 3). Surgery and interventional radiology were consulted and recommended the patient undergo immediate exploratory laparotomy. The patient received 2 units of packed red blood cells and fresh frozen plasma and was taken emergently to the operating room. Upon entering the abdomen, approximately 2.5 liters (L) of blood were noted predominantly along the left parametrial region with a distorted and friable appearance of the left adnexal structures. Profuse active bleeding was noted and suspected to originate from the left ovarian vessels which were successfully ligated in an effort to achieve hemostasis. The left ovary and fallopian tube were excised and sent to pathology. Continued bleeding was noted and the retroperitoneum was further explored. The left uterine artery was found to be actively bleeding and was ligated with hemoclips. Successful hemostasis was achieved with an estimated additional blood loss of 1.5L during exploration. On histopathology, a small focus of secretory type endometrium, suggestive of endometriosis, was identified in the excised left ovary and fallopian tube (Fig 4). Follow-up computed tomographic angiography confirmed successful ligation of the left uterine artery without additional active contrast extravasation (Fig 5). Follow-up fetal ultrasound demonstrated normal fetal heart tones with normal doppler wave form and a normal resistive index in the umbilical artery of 0.69 [reference range 0.60-0.87 at 22 weeks gestation]. The patient was discharged home 11 days post admission with instructions to follow-up in high risk OB/GYN clinic. The patient's pregnancy was further complicated by poor maternal weight gain and the fetus was noted to be small for gestational age, measuring in the 12th percentile. She was admitted for induction of labor at 36 weeks gestation and went on to deliver a 2346 g infant with Apgar scores of 8 and 9 via cesarean section.

DISCUSSION

Etiology & Demographics:

Spontaneous rupture of the uterine artery associated with pregnancy is extremely rare with approximately 100 cases reported in the literature [1]. Unfortunately, symptoms are often vague and maternal mortality is high, reported up to 30-40% [2]. As is our case, uterine rupture can occur in an otherwise uncomplicated pregnancy, and trends regarding trauma, gravidity, parity, age, or length of gestation have not been documented.

The exact etiology of spontaneous uterine artery rupture in relation to pregnancy is unknown, however pressure dynamics, anatomic and hormonal factors have been suggested [3]. Differential considerations include aneurysm formation, pseudo-aneurysm, congenital malformation, inflammatory or estrogen induced intimal changes of the vessel wall [4], local arterial erosion from endometriosis [5], or iatrogenic injury. In one case report, a non-pregnant woman with endometriosis was described as having local erosion of the uterine artery which resulted in spontaneous hemoperitoneum [5]. Although the patient was not pregnant at the time, this case underscores endometriosis as a causative agent in uterine artery rupture. A case of a 30-year-old G2P0 at 20 weeks gestation with history of endometriosis who presented with spontaneous hemoperitoneum due to uterine artery rupture has also been reported [6]. Given the histopathology in our case, as well as the absence of vascular malformations, underlying vascular disease, inflammatory conditions, or recent procedures, endometriosis should be considered as a possible etiology.

Differential Diagnoses:

The differential diagnosis of hemoperitoneum in a female of child-bearing age includes common gynecological conditions such ectopic pregnancy, ruptured ovarian cyst and endometriosis. In patients with a documented intrauterine pregnancy, hemoperitoneum has been reported in cases of uterine rupture, placenta percreta and placental abruption [7]. Vascular sources of hemoperitoneum should also be considered and include rupture of a visceral artery aneurysm. Splenic artery aneurysms in particular are more common in women and have increase rate of rupture during pregnancy [7].

Clinical & Imaging Findings:

Ultrasound is the first line imaging test in pregnant patients when available. It is the safest imaging modality with no adverse effects to the mother and fetus. Key ultrasound findings in the diagnosis of spontaneous extra-uterine hemorrhage in pregnancy include free intraperitoneal fluid in the setting of decreasing hemoglobin and hematocrit. These findings warrant additional radiographic analysis. Since it is fast, sensitive and readily available, contrast enhanced CT will be the imaging study performed in most cases. Although potential risks from ionizing radiation have been documented, maternal and fetal risks of delayed diagnosis in such emergencies outweigh these risks. The utilization of MRI in pregnancy avoids exposure to ionizing radiation and is preferred in some cases. However, it has limited utility in cases of vascular pathology since intravenous gadolinium contrast for MR angiography is an FDA class C agent and should not be used in pregnancy [8]. Relevant CT findings include hemoperitoneum and active contrast extravasation adjacent to the uterus or adnexa.

Treatment & Prognosis:

Patients with uterine artery rupture should be referred emergently to surgery and interventional radiology for discussion of therapeutic options. Maintaining hemodynamic stability should be of the utmost concern with appropriate blood transfusion and fluid replacement until the source of bleeding can be identified and appropriately managed. Though embolization may be considered a less invasive alternative to laparotomy, no studies have been conducted as to the preferential outcomes of embolization versus laparotomy in the unstable pregnant patient. However, all interventional procedures should take into consideration the hemodynamic stability of the patient and gestational age of the fetus, along with the availability of capable specialties and equipment. Studies comparing open surgery versus endovascular techniques have focused mainly on thoracic and abdominal aortic intervention; however, endovascular techniques have shown decreased operative time, length of hospital stay, and

blood loss without significant difference in mortality [9, 10, 11]. These reviews do not have significant evidence to support one modality over another and the risk/benefits stated should be well evaluated prior to procedure.

TEACHING POINT

Spontaneous rupture of a uterine artery in pregnancy, a rare OB/GYN emergency with high mortality, should be suspected in patients with unexplained large volume free intraperitoneal fluid and active contrast extravasation adjacent to adnexal structures. Patients should be emergently referred to obstetrics and gynecology, surgery, and interventional radiology for vessel ligation to protect both mother and fetal hemodynamic stability. Patient prognosis remains fair to good depending on rapid recognition of diagnosis and hemodynamic stability.

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FIGURES

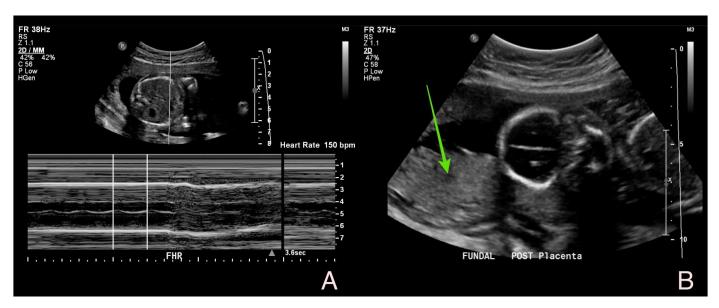


Figure 1: 36 year old pregnant female at 20 weeks gestation with ruptured uterine artery.

Findings: Initial transabdominal ultrasound demonstrates a live intrauterine gestation with normal fetal heart tones at 150 bpm and a normal homogenous placenta (B; green arrow).

Technique: Standard transabdominal ultrasound with gray scale and color doppler, 5-1MHz curved array C5-1 transducer

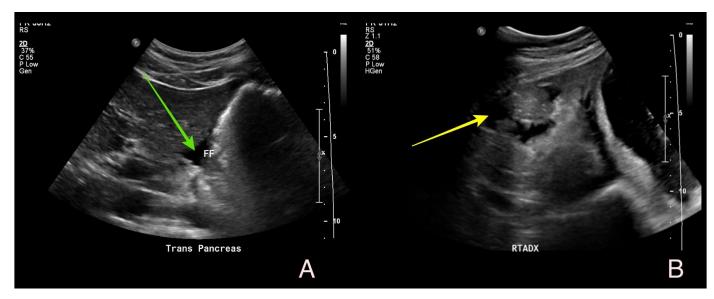


Figure 2: 36 year old pregnant female at 20 weeks gestation with ruptured uterine artery. Findings: Follow up transabdominal ultrasound demonstrates interval development of free fluid in the right upper quadrant (A; green arrow) and echogenic complex fluid in the right adnexa (B; yellow arrow)

Technique: Standard abdominal ultrasound with gray scale and color doppler, 5-2MHz curved array C5-2 transducer.

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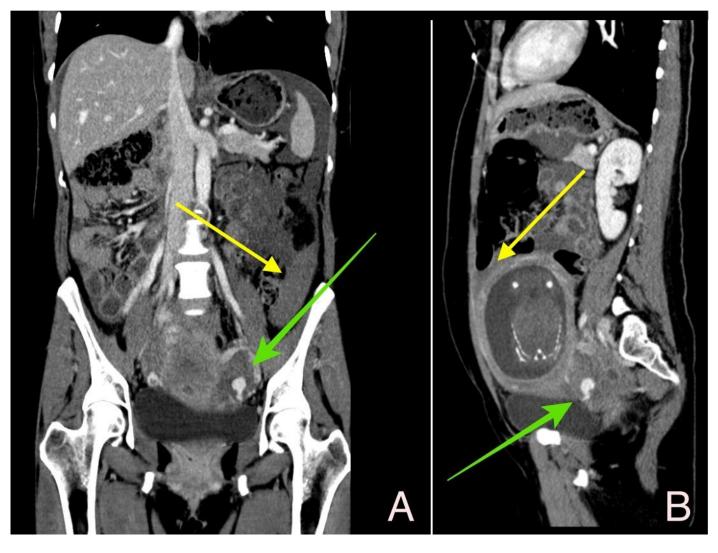


Figure 3: 36 year old pregnant female at 20 weeks gestation with ruptured uterine artery. Findings: Coronal (A) and Sagittal (B) reformations demonstrate large volume hemoperitoneum (A, B; yellow arrow) with an area of active contrast extravasation adjacent to the left adnexal vessels (A, B; green arrows). Technique: Contrast enhanced computed tomography of the abdomen and pelvis in portal venous phase. 120kV 182 mAs 100

Technique: Contrast enhanced computed tomography of the abdomen and pelvis in portal venous phase, 120kV, 182 mAs, 100 mL Optiray contrast, 3 mm slices.

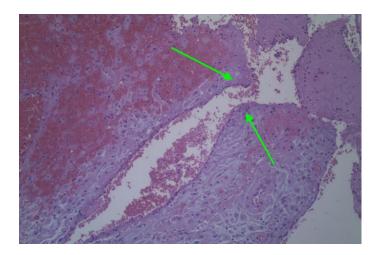


Figure 4 (left): 36 year old pregnant female at 20 weeks gestation with ruptured uterine artery. Findings: Decidualized stroma eroding into a vessel wall

consistent with endometriosis (green arrows). Technique: Standard Hemotoxylin and Eosin stain Obstetric & Gynecologic Radiology:

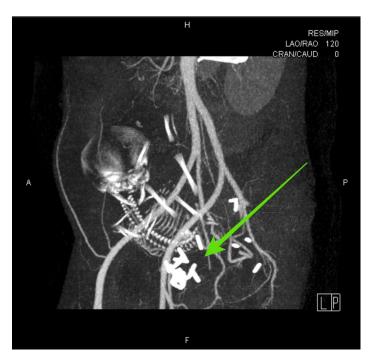


Figure 5 (left): 36 year old pregnant female at 20 weeks gestation with ruptured uterine artery.

Findings: 3D reformatted images demonstrate multiple surgical clips at the level of the left uterine artery without evidence of residual contrast extravasation (green arrow), intrauterine pregnancy is also visualized.

Technique: Contrast enhanced computed tomography angiograph of the abdomen and pelvis, 120 kV, 2330 mAs, 120 mL Optiray contrast, 1.5 mm slices

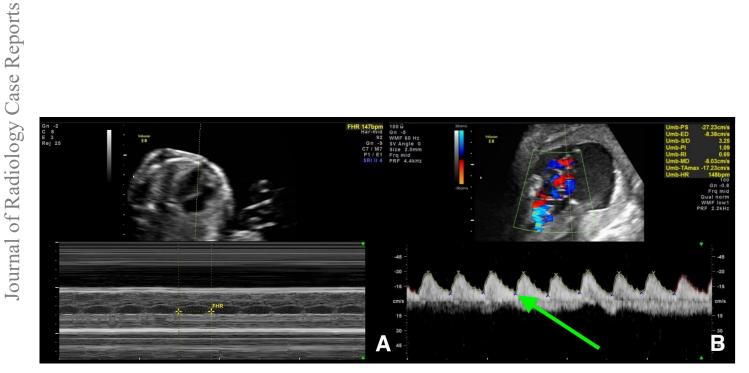


Figure 6: 36 year old pregnant female at 20 weeks gestation with ruptured uterine artery.

Findings: Ultrasound following uterine artery ligation demonstrated a viable intrauterine pregnancy with normal fetal heart rate at 147 bpm (A) and normal umbilical artery tracing (B) with normal diastolic flow (green arrow) and resistive index.

Technique: Standard abdominal ultrasound with gray scale and color doppler, 5-2MHz curved array C5-2 transducer.

Etiology	Exact etiology of spontaneous uterine artery rupture in relation to pregnancy is unknown; however possible considerations include aneurysm, pseudo-aneurysm, congenital malformation, inflammatory or estrogen induced intimal changes of the vessel wall, local arterial erosion from endometriosis, or iatrogenic injury.		
Incidence	Rare; approximately 100 reported cases in literature.		
Gender Ratio	Women only		
Age Predilection	Reproductive ages		
Treatment	Emergent laparotomy surgery		
	Interventional Radiology endovascular embolization		
Prognosis	Fair to Good, highly dependent on hemodynamic stability and early recognition.		
Findings on Imaging	Ultrasound - free intraperitoneal fluid		
	CT – Hemoperotineum, active contrast extravasation adjacent to the uterus or adnexa.		

Table 1: Summary table for spontaneous uterine artery rupture.

Differential Diagnosis	Ultrasound Findings	Computed Tomography Findings
Ruptured Uterine Artery	• Intraperitoneal fluid	 Hemoperitoneum Active contrast extravasation adjacent to the uterus or adnexa
Vascular Lesion (aneurysm or pseudoaneurysm)	 Aneurysm –Irregular vessel with abnormal flow dynamics on color Doppler, turbulent flow with aneurysmal sac. Pseudoaneurysm –Hypoechoic or anechoic structure adjacent to normal vessel, color flow demonstrates characteristic "ying-yang" sign, with swirling turbulent flow. Possible free intraperitoneal fluid 	 Aneurysm – widening of vessel wall with fusiform or sac-like appearance. Pseudoaneurysm—abnormal vessel contour communicating with the main vessel lumen. May have adjacent hematoma. If ruptured active contrast extravasation in peritoneal cavity
Ruptured Ovarian Cyst	• Anechoic or complex adnexal/ovarian mass with possible free fluid in pelvis.	• Adnexal Cyst containing low or high attenuating fluid depending on complexity. Free fluid may be visualized.
Ruptured Ectopic Pregnancy	• Possible adnexal mass and free blood in the peritoneal cavity.	• Extra-uterine heterogeneous mass with hemoperitoneum.
Placenta Percreta	 Placenta lacunae Irregular bladder wall with vascularity Loss of retroplacental clear space Decreased myometrial thickness (<1mm) 	• No role for initial evaluation.
Placenta Abruption	Possible retroplacental hematoma	• No role for initial evaluation.

Table 2: Differential Diagnosis table for nontraumatic hemoperitoneum in gravid and child bearing aged female.

ABBREVIATIONS

CBC = Complete Blood Count cm = Centimeter CT = Computed Tomography CTA = Computed Tomographic Angiography dL = Deciliter gm = Gram HBG = Hemoglobin L= Liter OB/GYN = Obstetrics and Gynecology UL = Microliter US = Ultrasound WBC = White Blood Count

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Uterus; Uterine artery; Rupture; Hemorrhage; Spontaneous