

Emergency Radiology

Uterine Artery Pseudoaneurysm in the Setting of Deep Endometriosis: An Uncommon Cause of Hemoperitoneum in Pregnancy

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Corresponding Author:	Zoe McGovern Feld, B.A. Florida International University Herbert Wertheim College of Medicine Miami, UNITED STATES
Corresponding Author Secondary Information:	
Corresponding Author's Institution:	Florida International University Herbert Wertheim College of Medicine
Corresponding Author's Secondary Institution:	
First Author:	Zoe Feld, B.A.
First Author Secondary Information:	
Order of Authors:	Zoe Feld, B.A. Tami Rowen, M.D. Andrew Callen, M.D. Ruth Goldstein, M.D. Liina Poder, M.D.
Order of Authors Secondary Information:	
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Abstract:	<p>Uterine, ovarian, and placental pathologies are among the differential considerations for a pregnant woman presenting with abdominal and pelvic pain. Imaging plays a key role in the initial workup of these patients. Sonography is often the first line test, however evaluation of pelvic pathology can be limited in the gravid state, especially in mid- or late- term pregnancy. We present a case of a pregnant woman who came to the emergency room at 25 weeks with acute abdominal and pelvic pain. Both ultrasound and MR imaging findings revealed intraperitoneal hemorrhage, initially of unknown origin, as well as endometriomas and deep endometriosis. Only postpartum imaging confirmed a uterine artery pseudoaneurysm (PSA) presumably due to decidual reaction in deep endometriosis. We speculate the intraperitoneal hemorrhage was subsequently due to the PSA. This case demonstrates that if hemorrhage is not recognized promptly, it can lead to hemodynamic instability, as well as premature labor and delivery.</p>



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Abstract:

Uterine, ovarian, and placental pathologies are among the differential considerations for a pregnant woman presenting with abdominal and pelvic pain. Imaging plays a key role in the initial workup of these patients. Sonography is often the first line test, however evaluation of pelvic pathology can be limited in the gravid state, especially in mid- or late- term pregnancy. We present a case of a pregnant woman who came to the emergency room at 25 weeks with acute abdominal and pelvic pain. Both ultrasound and MR imaging findings revealed intraperitoneal hemorrhage, initially of unknown origin, as well as endometriomas and deep endometriosis. Only postpartum imaging confirmed a uterine artery pseudoaneurysm (PSA) presumably due to decidual reaction in deep endometriosis. We speculate the intraperitoneal hemorrhage was subsequently due to the PSA. This case demonstrates that if hemorrhage is not recognized promptly, it can lead to hemodynamic instability, as well as premature labor and delivery.

Background:

Uterine, ovarian, and placental pathologies are among the differential considerations for a pregnant woman presenting with abdominal and pelvic pain. Imaging plays a key role in the initial workup of these patients. Sonography is often the first line test, however evaluation of pelvic pathology can be limited in the gravid state, especially in mid- or late- term pregnancy. In these cases, magnetic resonance imaging (MRI) can be helpful but may also be difficult to interpret in advanced stages of pregnancy. Endometriosis-associated spontaneous hemoperitoneum in pregnancy (SHiP) is a very rare condition and is difficult to diagnose preoperatively. Patients may be initially hemodynamically stable with a nonspecific physical exam.¹ Complications of endometriosis, specifically vascular compromise, are not common enough to be routinely considered in the patient's initial workup but the following case highlights the importance of this diagnosis. We present a case of a pregnant woman who came to the emergency room at 25 weeks with acute abdominal and pelvic pain. Both ultrasound and MR imaging findings revealed intraperitoneal hemorrhage, initially of unknown origin, as well as endometriomas and deep endometriosis. Only postpartum imaging confirmed a uterine artery pseudoaneurysm (PSA) presumably due to decidual reaction in deep endometriosis. We speculate the intraperitoneal hemorrhage was subsequently due to the PSA. We found no previously reported cases in the literature of spontaneous PSA developing in decidualized endometriosis. Intraperitoneal hemorrhage is a serious complication during pregnancy. This case demonstrates that if hemorrhage is not recognized promptly, it can lead to hemodynamic instability, as well as premature labor and delivery.

Case presentation:

A 37-year-old G1P0000 woman at 23w2d was admitted for significant abdominal pain. She had a past medical history significant for chronic microcytic anemia and granulomatosis with polyangiitis (GPA), placental mosaicism and intrauterine growth restriction as well as a bladder mass noted on routine obstetric ultrasound, felt to be benign on cystoscopy (and turned out to be another site of deep endometriosis). On labor and delivery, she was found to have frequent contractions on tocometer. Her exam was unremarkable but her laboratory workup showed a

hemoglobin was noted to drop from 11.5 10 days prior to 8.9 on the day of admission. A limited obstetric ultrasound showed no evidence of placental abruption. She had no cervical change and was presumed to have pain from preterm contractions, was placed on disability due to the pain and followed up as an outpatient. She was seen for a formal obstetric ultrasound 11 days after discharge, now 25w1d and was continuing to report severe abdominal pain, reporting 5 days of acute on chronic sharp right lower quadrant abdominal pain. There was associated anorexia without nausea, vomiting, vaginal bleeding or discharge. On exam she was afebrile and without peritoneal signs. Due to concern for ovarian torsion, pelvic ultrasound was performed, in which the ovaries were not adequately visualized, and multiple dilated veins were seen in the left adnexal region (**Figure 1**). MRI without contrast was recommended to exclude ovarian torsion and look for other potential causes of pelvic pain. MRI was limited due to the patient's discomfort during the exam but revealed extrauterine locules of T1 hyperintense material mostly surrounding left adnexa and posterior cul-de-sac in keeping with intraperitoneal hemorrhage. (**Figure 2**) The left ovary had no signs of torsion but contained a complex cyst with internal blood products, thought to represent hemorrhagic cyst versus an endometrioma. Incidentally noted was left uterosacral ligament asymmetric thickening with heterogeneous T2 signal with internal hyperintense foci on T1 (**Figure 3**). Asymmetric hydronephrosis was present on the left side with narrowing of the distal ureter likely due to endometriosis.

The patient was admitted for pain control and diagnostic workup. Her pain worsened and during the observation, she reported a sudden and worsening pain, and an increase in uterine contractions with cervical change. Repeat labs showed her hemoglobin dropped from 8.7 to 6.3, requiring transfusion of two units of packed red blood cells. Bedside ultrasound revealed large volume free fluid in her abdomen. The patient was taken for diagnostic laparoscopy, during which 1600 cc of organized blood was evacuated from the pelvis. The left ovarian lesion was identified that was moderately bleeding, intraoperatively thought to represent a hemorrhagic cyst or endometrioma; peritoneal biopsies were obtained and sent for pathologic analysis, which subsequently demonstrated decidual reaction. On post-operative day 1, the patient experienced preterm premature rupture of membranes and delivered via spontaneous vaginal delivery at 25wk6d. The neonate expired at four days of life. The patient's clinical status stabilized post-partum and she was discharged from the hospital with a presumed diagnosis of endometriosis.

Follow up MRI with contrast three weeks after discharge revealed deep endometriosis involving the left pelvic sidewall and left uterosacral ligament, decreased in extent compared to her prior intra-partum exam. Within the region of deep endometriosis was a focus of intense enhancement measuring 11 mm, suggestive of pseudoaneurysm. CT angiogram of the abdomen and pelvis delineated the pseudoaneurysm deriving from a branch of the left uterine artery, measuring 14 x 9 mm and with no evidence of active extravasation. Subsequent embolization attempt by interventional radiology confirmed the pseudoaneurysm but embolization was unsuccessful due to uterine artery spasm. (**Figure 4**) Two weeks later, repeat MRI showed resolution of the pseudoaneurysm. Patient is currently being followed for treatment of her newly diagnosed deep endometriosis with the goal of conceiving in the near future and carrying to term.

Discussion:

When a pregnant patient presents with lower abdominal or pelvic pain, transabdominal sonography should be performed as the initial imaging modality. As was the case with this patient, peritoneal implants in endometriosis can often be difficult to visualize with ultrasound.² If the pathologic process is incompletely characterized, as in this case, it is reasonable to proceed to noncontrast MRI, which is more specific for the diagnosis of endometriosis.³

Deep endometriosis is defined as subperitoneal invasion by endometriotic lesions causing reactive fibrous changes which typically present as T2 hypointense masses or nodules with irregular, indistinct or stellate margins. Intermingled T2 hyperintense foci may be observed, reflecting ectopic endometrial glands. There may or may not be T1 hyperintense internal endometriotic foci. The fibrous and reactive changes may demonstrate enhancement on post contrast imaging and may raise concern for a possible neoplastic process. Typical anatomic locations include the rectovaginal septum, uterosacral ligaments, vesicovaginal septum, vesicouterine pouch, prevesical space, alimentary tract, and urinary tract. Involvement of the urinary tract or uterosacral ligaments can lead to ureteral strictures and hydronephrosis. The presence of these characteristic signal characteristics in the typical anatomic locations are diagnostic of deep endometriosis.⁴

The natural history of endometriosis involves a gradual decrease in symptoms and imaging findings throughout pregnancy, with the notable exception of the phenomenon of decidualization. With decidual reaction induced from the gravid state, neovascularity can develop. In this case, we speculate decidualization and subsequent neovascularity led to the eventual development of pseudoaneurysm.⁵

Uterine artery pseudoaneurysm is a rare complication of endometriosis. Ferrero et al. described uterine artery pseudoaneurysm after excision of pelvic endometriosis, but no case has been reported in the literature of a pseudoaneurysm arising from decidualized endometriosis without previous surgery or an inciting event.⁶ Although no cases have been reported of primary uterine artery aneurysm secondary to GPA, it is possible that this patient's condition predisposed her to vascular injury.

In this case, secondary arterial pseudoaneurysm was not detected until post partum contrast enhanced MRI. Gadolinium is generally avoided in pregnancy due to its ability to cross the placenta and potentially harm the fetus. The American College of Obstetrics and Gynecology recommends the use of MRI contrast should be limited to situations where the diagnostic benefits clearly outweigh the possible risks.⁷ Although difficult to determine prospectively, in this case the risk from complications of intraperitoneal hemorrhage outweighs the potential risk from gadolinium administration. We postulate that if contrast imaging was obtained earlier, the PSA would have been detected and treated sooner, and premature delivery potentially could have been avoided.

In summary, understanding and utilizing ultrasound and MRI as complementary modalities in the pregnant patient with abdominal pain can expedite the diagnosis and lead to expedited treatment and preservation of pregnancy. This case highlights a rare but important complication of endometriosis in pregnancy.

Conflict of Interest: The authors declare that they have no conflict of interest.

Informed Consent: For this type of study, formal consent is not required.

References

1. Leone Roberti Maggiore U, Ferrero S, Mangili G et al. A systematic review on endometriosis during pregnancy: diagnosis, misdiagnosis, complications and outcomes. *Human Reproduction Update*. 2015;22(1):70-103.
2. Olive DL, Schwartz LB. Endometriosis. *N. Engl. J. Med.* 1993;328 (24): 1759-69
3. Woodward PJ, Sohaey R, Mezzetti TP. Endometriosis: radiologic-pathologic correlation. *Radiographics*. 21 (1): 193-216
4. Coutinho et al . MR Imaging in Deep Pelvic Endometriosis: A pictorial Essay. *RadioGraphics* 2011; 31:549–567
5. Poder L, Coakley FV, Rabban JT, Goldstein RB, Aziz S, Chen LM. Decidualized endometrioma during pregnancy: recognizing an imaging mimic of ovarian malignancy. *J Comput Assist Tomogr.* 2008;32(4):555–8.
6. Ferrero et al. Unusual complication of excision of pelvic endometriosis: pseudoaneurysm of the left uterine artery. *Fertil Steril.* 2010 Jan;93(1):264-6
7. Guidelines for diagnostic imaging during pregnancy and lactation. Committee Opinion No. 656. American College of Obstetricians and Gynecologists. *Obstet Gynecol* 2016;127:e75–8

Figures

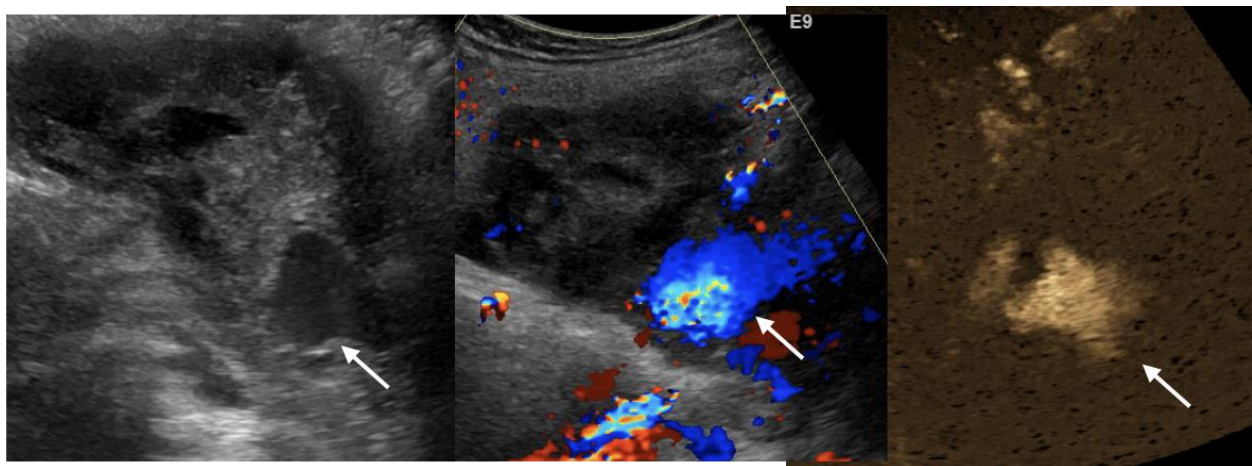


Figure 1: Initial gray scale (A) duplex US (B) and B-mode (C) did not clearly visualize adnexa. Dilated pelvic veins were described which were thought to represent physiologic changes. Only retrospectively was a focal areal of increased vascularity identified correlating to the PSA on postpartum MRI and CTA. (arrows)

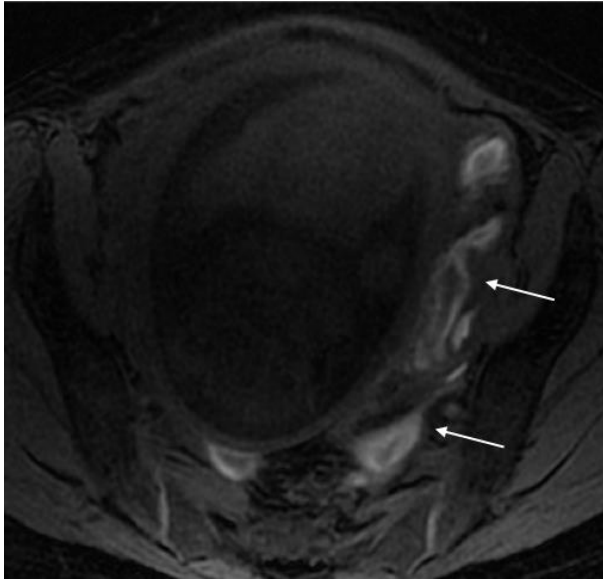


Figure 2: A. T1 axial image demonstrating hyper intense material surrounding left adnexa and cut de sac (arrow), representing extra uterine blood products consistent with intraperitoneal hemorrhage.

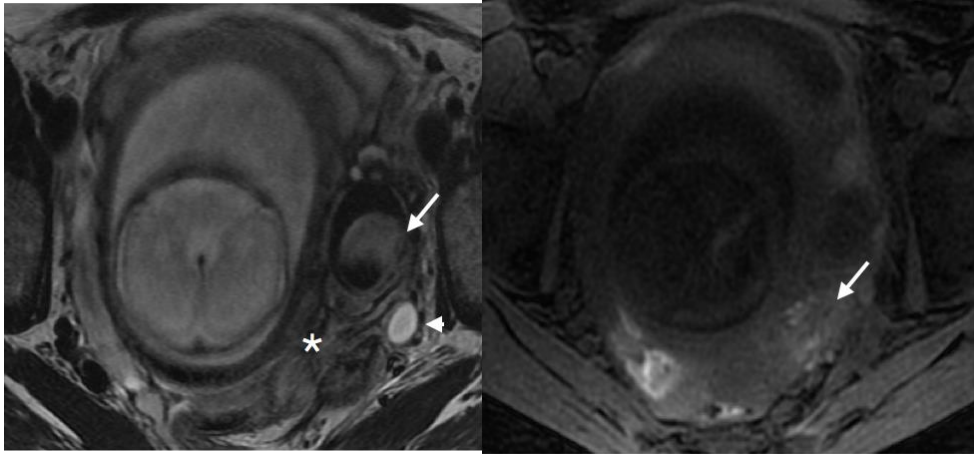


Figure 3: A. T2 axial image demonstrated possible hemorrhagic cyst versus endometrioma (arrow). Incidentally noted is asymmetric thickening of left uterosacral ligament (asterisk) and secondarily dilated left ureter (arrow head). B. T1 hyperintense foci in the thickened left uterosacral ligament (arrow). The constellation of findings on T1 and T2 are strongly suggestive of deep endometriosis involving the left uterosacral ligament.

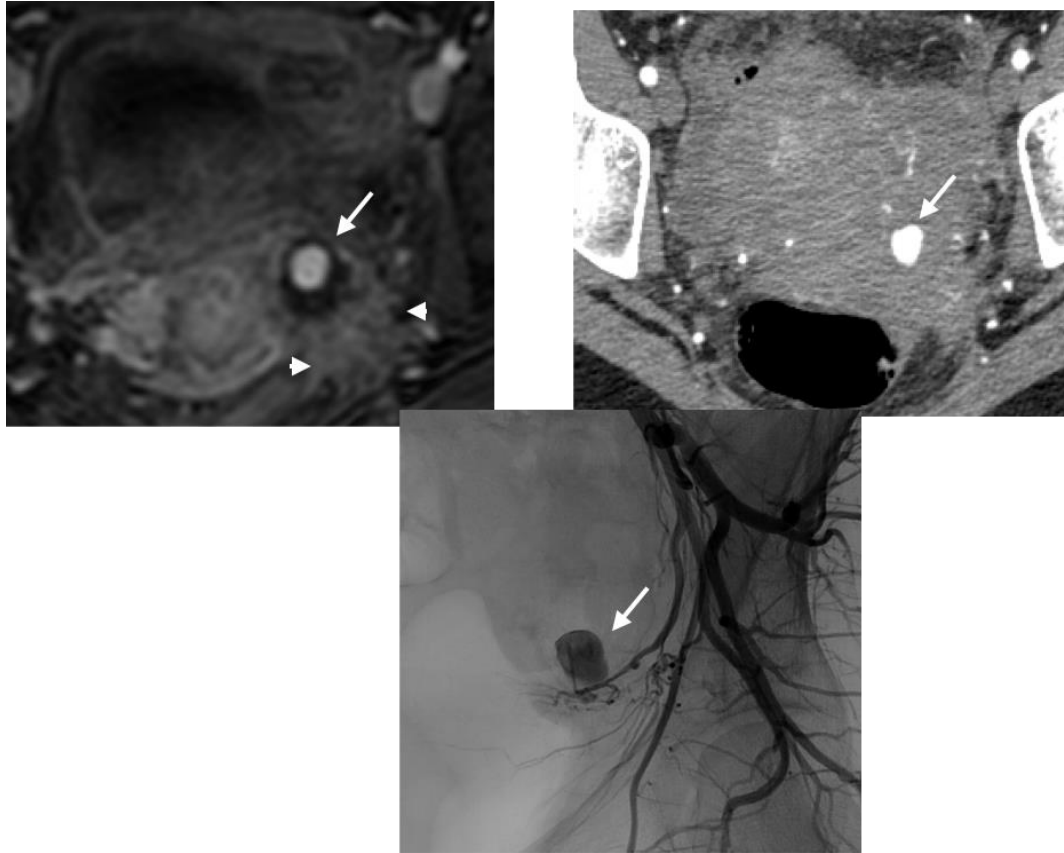


Figure 4: A. T1 post gadolinium image demonstrates mildly enhancing thickened left uterosacral ligament (arrowheads) with focal rounded area of brisk enhancement (arrow). B. Branch uterine artery PSA was confirmed on subsequent CT. C. Pre-embolization angiogram concurred with the cross-sectional imaging findings (arrow)