Case Report

Multimodality Imaging for Rare Presentation of Placenta Increta in the First Trimester in a Patient with Previous Cesarean Section and Asherman Syndrome

Harshna V. Vadvala[1]*, Jonathan E. Henning[2], Abimbola Aina-Mumuney[3]

[1]Department of Radiology, Moffitt Cancer Center, University of South Florida, Tampa, FL, USA, [2]University of South Florida Morsani College of Medicine, Tampa, Fl, USA, [3]Division of Maternal-Fetal Medicine, Department of Gynecology/Obstetrics, Johns Hopkins University School of Medicine, Baltimore, MD, USA

Abstract

We report multi-modality imaging (Ultrasound and Magnetic Resonance Imaging) findings of a rare complication in a multi-gravida patient with history of Asherman syndrome presenting with placenta increta in a cesarean scar ectopic pregnancy. The appropriate diagnosis was established with imaging and patient was managed surgically with total abdominal hysterectomy and bilateral salpingectomy. Asherman syndrome and its management of hysteroscopic adhesiolysis are associated with increased odds of placenta accreta spectrum and postpartum hemorrhage. Patients with Asherman syndrome are considered high risk in pregnancy and should be closely monitored for placental site abnormalities during current and subsequent pregnancies.

Keywords: Asherman syndrome, cesarean scar ectopic pregnancy, multi-modality imaging, placenta increta

Introduction

Imaging is critical for diagnosing a first trimester case of placenta increta in a cesarean scar ectopic pregnancy in patients with Asherman syndrome. Suspicion for the placenta accreta spectrum (PAS) must be high when confronted with pregnant women with history of uterine surgery, Asherman syndrome, placenta previa and/or assisted reproduction techniques. This case presents images that may guide sonologists/radiologists who encounter this scenario in early gestation.

Literature describes Asherman syndrome's association with placenta accreta, but the diagnosis is often made through pathologic specimens and has not been often depicted via two imaging modalities with the correlation between ultrasonography and magnetic resonance imaging (MRI) findings.

Case Report

A pregnant 36-year-old patient (G6, P2-0-3-2) with a history of Asherman syndrome and prior cesarean section initially presented to an affiliated community hospital for heavy vaginal bleeding, crampy abdominal pain, and syncope. Ultrasound (US) imaging suggested placenta accreta; therefore, she was referred to our tertiary care center for further management. The patient had 2 full term deliveries (a cesarean section followed by a vaginal birth after cesarean) prior to two dilation and suction curettage procedures (the first after an incomplete abortion and the second to remove scar tissue secondary to Asherman syndrome).

Initial US demonstrated single intrauterine pregnancy (IUP) (CRL of 6w6d) with cardiac activity and abnormal findings in the lower uterine segment. Our maternal and fetal medicine department evaluated the patient further, where subsequent imaging showed a collapsed, unexpanded endometrial cavity in the uterine fundus superior to the gestational sac (GS), in keeping with the prior diagnosis of Asherman

Received: 29-03-2023 **Revised:** 27-04-2023 **Accepted:** 18-05-2023 **Available Online:** 28-08-2024

Access this article online

Quick Response Code:

Website: https://journals.lww.com/jmut

DOI: 10.4103/jmu.jmu_33_23

Address for correspondence: Dr. Harshna V. Vadvala, Department of Radiology, Moffitt Cancer Center, University of South Florida, 12902 USF Magnolia Drive, Tampa, FL 33612, USA.
E-mail: drharshna@gmail.com

How to cite this article: Vadvala HV, Henning JE, Aina-Mumuney A. Multimodality imaging for rare presentation of placenta increta in the first trimester in a patient with previous cesarean section and Asherman syndrome. J Med Ultrasound 2024;32:262-5.

syndrome [Figure 1]. Multiple tiny anechoic cystic spaces representing placental lacunae were seen in the lower uterine segment adjacent to the site of the presumed IUP [Figure 1]. Color Doppler showed increased vascularity within these lacunar spaces on the periphery of the GS, particularly along the anterior lower uterine wall. Short interval follow-up US yielded no cardiac activity at 8 weeks gestation suggestive of failed pregnancy at which point the patient reported sharp and severe pelvic cramps. US showed placental lacunae, abnormal uteroplacental interface, absence of retroplacental clear space with retroplacental loss of myometrium. Swirling sign was seen on Doppler. The findings were suspicious for PAS [Figure 2].[1] Patient was therefore transferred to our tertiary care hospital.

On admission, MRI of the pelvis confirmed a gravid uterus with heterogeneously hyperintense T2 signal in the lower uterine segment and upper cervix where the GS, embryo and placenta were not discernible as separate structures compatible with failed early pregnancy [Figure 3]. An enhancing, infiltrative, complex, solid, and cystic mass-like lesion was seen invading the entire myometrium of the anterior lower uterine segment and upper cervix [Figure 4]. Placenta previa was also present. There was the presence of a focal lumpy contour of the uterus, T2 dark intraplacental bands, focal gap in myometrium and heterogenous placenta signal, which was suggestive of MR signs of invasive placentation.[2] There was tenting with scar defect seen at the anterior surface of the low uterine segment in keeping with the history of prior cesarean sections which suggested cesarean scar pregnancy. MRI differential included placenta accreta complex with cesarean scar ectopic pregnancy and less likely gestational trophoblastic neoplasm (partial mole). Beta human chorionic gonadotropin (hCG) levels ranged between 10,736 and 61,736 mIU/mL over a month's time.

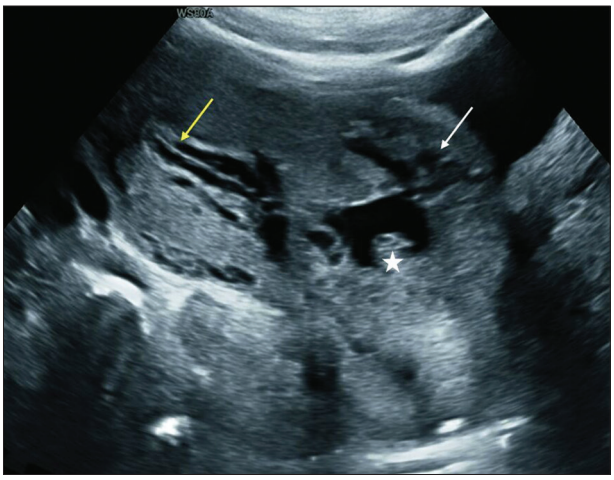

Figure 1: Gray scale US at 8 gestational weeks shows unexpanded endometrium in the uterine fundus superior to the IUP (white star), a sequela of known Asherman syndrome (yellow arrow). Multiple tiny anechoic cystic spaces in the lower uterine segment adjacent to the site of IUP representing placental lacunae (white arrow). US: Ultrasound, IUP: Intrauterine pregnancy

Urgent total abdominal hysterectomy and bilateral salpingectomy were performed after bilateral ureteral stent placement. The patient verbalized no desire for future fertility but requested retention of her cervix for her sexual health. She was counselled preoperatively that this may not be feasible, given the level of involvement of the cervix on imaging. The patient required an infusion of packed red blood cells and Plasma-Lyte intraoperatively. Histopathology of the surgical specimen showed chorionic villi and intermediate trophoblast extending to the outer myometrium with intermediate trophoblast also extending into extrauterine soft tissue; however, chorionic villi were limited to the myometrium without definitive extrauterine extension consistent with placenta increta involving the lower uterine segment and endocervix [Figure 5]. The remaining uterus showed decidualized endometrium and adenomyosis.

The villous cytotrophoblast and stromal cells were positive for p57 [Figure 6], which in conjunction with the morphology, ruled out the possibility of a complete hydatidiform mole (including its early form). There were no postoperative complications.

Discussion

We present a rare, first-trimester case with multi-modality imaging and pathological findings of placenta increta in a cesarean scar ectopic pregnancy in a patient with Asherman syndrome and adenomyosis.

Abnormal placental attachment to the uterine wall and related complexities are referred to as PAS. In placenta *accreta*, the villi are adherent to the myometrial surface without intervening decidua, in *increta*, the villi extend further into the myometrium, and in *percreta*, the villi penetrate the entire uterine wall, including serosa.[3] Identified risk factors for PAS include smoking, advanced maternal age, uterine anomalies, prior uterine surgery, Asherman syndrome, and the use of assisted reproductive technologies.[4]

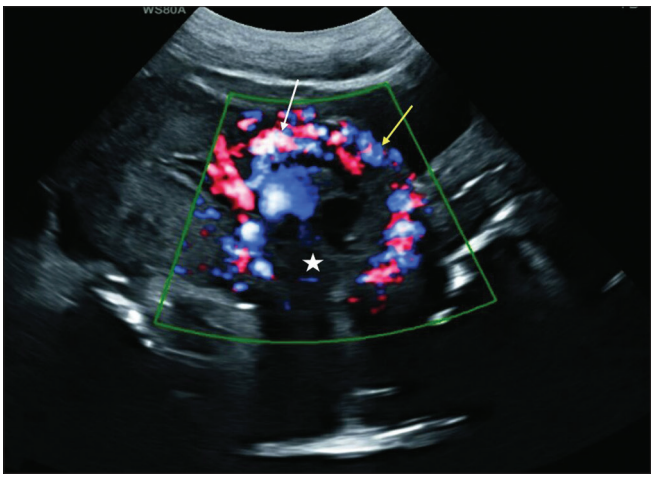

Figure 2: Color doppler US at 8 gestational weeks shows increased vascularity within lacunar spaces (white arrow) on the periphery of the IUP (white star) particularly along the anterior uterine wall. Placenta appeared to protrude into the uterine musculature (yellow arrow). US: Ultrasound, IUP: Intrauterine pregnancy

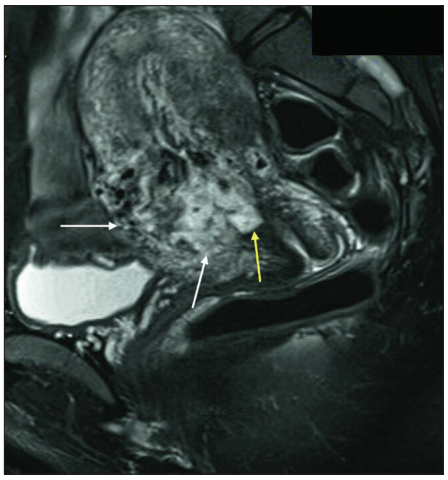

Figure 3: MRI pelvis T2 weighted sequence at 8 gestational weeks shows irregular mass-like lesion with cystic lesions invading entire thickness of anterior lower uterine segment and upper cervix (white arrows). No definitive GS, embryo and placenta seen as separate structures, compatible with failed early pregnancy. Mild placenta previa (yellow arrow). MRI: Magnetic resonance imaging, GS: Gestational sac

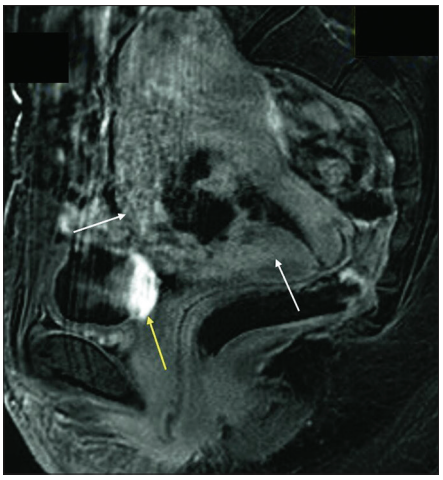

Figure 4: MRI pelvis T1 weighted sequence at 8 gestational weeks with gadolinium contrast (subtraction image) shows enhancing, infiltrative, complex solid cystic mass-like lesion invading the entire thickness of myometrium of the anterior lower uterine segment and upper cervix (white arrows). Urinary bladder remained intact (yellow arrow). MRI: Magnetic resonance imaging

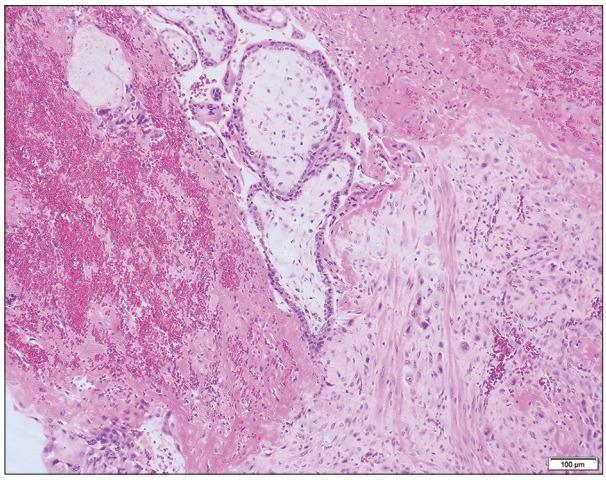

Figure 5: H and E staining of chorionic villi extending to the outer myometrium. H and E: Hematoxylin and eosin 100X

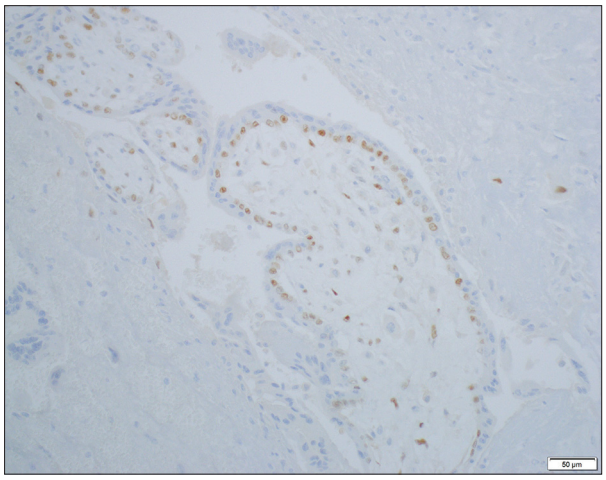

Figure 6: There is presence of p57 staining in villous cytotrophoblast and stromal cells 200X

This patient, in particular, had multiple risk factors for PAS, including placenta previa, prior cesarean section, Asherman syndrome, and (although not known preoperatively) adenomyosis.[5] It is difficult to determine which of these played the greatest role in her outcome, but perhaps the combination led to an earlier, more extreme presentation of PAS with involvement of nearly the entire uterine wall by 8 weeks gestation. To our knowledge, this is the first case of placenta accreta with this unique combination of conditions at this early gestational age.

In Asherman syndrome, there is partial or complete obstruction of the endometrial cavity due to damage to the basal layer of the endometrium, which could be caused by intrauterine procedures and infections often associated with a failed pregnancy.[6] The hysteroscopic adhesiolysis procedure used to manage Asherman syndrome is known to be associated with retained placenta, adherent placenta, placenta accreta, placenta previa, and postpartum hemorrhage in subsequent pregnancies.[7]

In this patient, radiographic investigation began with the US, followed by MRI. The key US findings in PAS are placental lacunae, abnormal color Doppler imaging patterns, loss of the retroplacental clear space, and reduced myometrial thickness. Placental lacunae are tiny hypervascular cystic-appearing structures with turbulent flow in the placental parenchyma, displaying "moth-eaten" or "Swiss cheese" appearance on US. The presence of multiple placental lacunae has the highest sensitivity and is the most predictive sign for PAS.[8] The absence of the retroplacental hypoechoic line, which is usually present with normal placentation, has been described for PAS.[1] If found during sonographic imaging of high-risk pregnancies, these findings should prompt further investigation.

MRI with gadolinium was utilized as a problem-solving modality in this case because the pregnancy was no longer viable, but MRI without contrast may also be illuminating. The key findings are abnormal focal uterine bulging at the site of placental invasion, abnormal heterogeneous T2 signal intensity within the placenta, absent myometrial signal underneath the placenta site, hypointense intraplacental bands on T2-weighted images and hyperenhancing invasive mass lesions within the myometrium on gadolinium imaging.[9] The uneven or random distribution of abnormal intraplacental bands with variable thickness extending from the uterine-myometrial interface may represent a reliable sign for PAS.

Gestational trophoblastic neoplasm was a close differential in this case given the hypervascular mass-like lesion invading the myometrium; however, clinical presentation and lower beta-hCG levels favoured placenta increta in cesarean scar ectopic pregnancy.[10]

Asherman syndrome and its association with placenta accreta has been described in case reports and small case series,[11-13] but the diagnosis is often noted on pathologic specimens and has seldom been depicted via two different imaging modalities with a correlation between US and MRI findings.

Total hysterectomy and bilateral salpingectomy were selected to achieve complete resolution in this case because the patient had completed her childbearing. Certainly, more conservative methods of treatment are available for those desiring to preserve future fertility such as uterine artery embolization (UAE). UAE may not be the best choice for patients who already have Asherman syndrome though, as this procedure has been shown to cause severe intrauterine adhesions.[14]

This case demonstrates the utility of multi-modality imaging even at extremely early gestational ages when a high clinical index of suspicion warrants their use.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that her name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

References

1. Abinader RR, Macdisi N, El Moudden I, Abuhamad A. First-trimester ultrasound diagnostic features of placenta accreta spectrum in low-implantation pregnancy. Ultrasound Obstet Gynecol 2022;59:457-64.
2. Kilcoyne A, Shenoy-Bhangle AS, Roberts DJ, Sisodia RC, Gervais DA, Lee SI. MRI of placenta accreta, placenta increta, and placenta percreta: Pearls and pitfalls. AJR Am J Roentgenol 2017;208:214-21.
3. Jauniaux E, Collins S, Burton GJ. Placenta accreta spectrum: Pathophysiology and evidence-based anatomy for prenatal ultrasound imaging. Am J Obstet Gynecol 2018;218:75-87.
4. Badr DA, Al Hassan J, Salem Wehbe G, Ramadan MK. Uterine body placenta accreta spectrum: A detailed literature review. Placenta 2020;95:44-52.
5. Mandelbaum RS, Melville SJ, Violette CJ, Guner JZ, Doody KA, Matsuzaki S, *et al.* The association between uterine adenomyosis and adverse obstetric outcomes: A propensity score-matched analysis. Acta Obstet Gynecol Scand 2023. p. 1-10. [doi: 10.1111/aogs.14581].
6. Smikle C, Yarrarapu SN, Khetarpal S. Asherman syndrome. In: StatPearls. Treasure Island (FL): StatPearls Publishing; 2023. Available from: https://www.ncbi.nlm.nih.gov/books/NBK448088. [Last accessed on 2023 Mar 20].
7. Xiao S, Wan Y, Xue M, Zeng X, Xiao F, Xu D, *et al.* Etiology, treatment, and reproductive prognosis of women with moderate-to-severe intrauterine adhesions. Int J Gynaecol Obstet 2014;125:121-4.
8. Comstock CH. Antenatal diagnosis of placenta accreta: A review. Ultrasound Obstet Gynecol 2005;26:89-96.
9. Lax A, Prince MR, Mennitt KW, Schwebach JR, Budorick NE. The value of specific MRI features in the evaluation of suspected placental invasion. Magn Reson Imaging 2007;25:87-93.
10. Shaaban AM, Rezvani M, Haroun RR, Kennedy AM, Elsayes KM, Olpin JD, *et al.* Gestational trophoblastic disease: Clinical and imaging features. Radiographics 2017;37:681-700.
11. Engelbrechtsen L, Langhoff-Roos J, Kjer JJ, Istre O. Placenta accreta: Adherent placenta due to A Sherman syndrome. Clin Case Rep 2015;3:175-8.
12. Dmowski WP, Greenblatt RB. Asherman's syndrome and risk of placenta accreta. Obstet Gynecol 1969;34:288-99.
13. Sonan Y, Aoki S, Enomoto K, Seki K, Miyagi E. Placenta accreta following hysteroscopic lysis of adhesions caused by Asherman's syndrome: A case report and literature review. Case Rep Obstet Gynecol 2018;2018:6968382.
14. Jegaden M, Bleas C, Debras E, Couet D, Pourcelot AG, Capmas P, *et al*. Asherman syndrome after uterine artery embolization: A cohort study about surgery management and fertility outcomes. J Minim Invasive Gynecol 2023;30:494-501.