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Case Report

Uterine window and placenta accreta spectrum in a dichorionic, diamniotic twin gestation complicated by postpartum hemorrhage ☆

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ABSTRACT

Cesarean scar defects encompass a spectrum of separation of the uterine layers that increase risk of uterine rupture during labor and delivery. When coupled with the presence of placenta accreta spectrum, a condition characterized by abnormal adherence to or invasion of the myometrium by the placenta, the risk of life-threatening hemorrhage significantly increases. As rates of cesarean delivery increase, it is important to understand the imaging findings typical of both conditions. We present the case of a 30-year-old patient with a dichorionic, diamniotic twin gestation complicated by uterine window and placenta accreta spectrum.

Introduction

Cesarean section (CS) is a surgical means of delivering a fetus when vaginal delivery is not possible. Rates of CS are increasing worldwide, currently estimated at 21% and projected to reach a global rate of 29% in 2030 [1,2]. Several long-term complications are associated with CS including cesarean scar defects (CSDs), which may manifest along a spectrum of myometrial tissue compromise at the location of prior hysterotomy incision repair. The most feared of these is uterine rupture, which is defined as complete separation of the 3 layers of the uterus: the endometrium (inner epithelial layer), myometrium (smooth muscle layer) and outer serosal layer, usually occurring acutely during labor and resulting in clinically significant hemoperitoneum and hemodynamic compromise [3]. Uterine dehiscence (also referred to as uterine "window") is defined as incomplete separation of the myometrium with an intact overlying serosa and is usually clinically occult and diagnosed incidentally at repeat cesarean birth (or during ultrasound), without maternal or neonatal adverse effect [4]. In the setting of uterine dehiscence or

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near dehiscence, imaging may reveal a uterine "window," through which the fetus can be visualized under a layer of markedly thinned myometrium and/or absent myometrium with intact overlying serosa. CSDs are important to consider in multiparous patients, especially those considering a trial of labor after cesarean (TOLAC), due to increased risk of uterine dehiscence and rupture. Rates of attempted TOLACs are also increasing, from 15.3% in 2010 to 21.7% in 2020 [5].

Prior CS is also a major risk factor for placenta accreta spectrum (PAS), which is defined as abnormal adherence of the placenta to the myometrium and/or myometrial invasion [6]. PAS also occurs along a spectrum which includes placenta accreta, increta and percreta. Other risk factors for PAS include advanced maternal age, placenta previa, previous uterine instrumentation including dilation and curettage (D&C) and assisted reproductive technologies (ART). The main concern in cases of PAS is the potential for severe peripartum hemorrhage, which may require emergency hysterectomy [7]. The incidence of PAS is rising worldwide, increasing in incidence from 1 in 2510 women before 1994 to 1 in 272 women in 2016 [8,9]. Placenta previa, which describes placental implantation over the internal cervical os, frequently co-exists with PAS due to tethering of the placenta in the lower uterine segment secondary to scarring. In patients with placenta previa, the risk of placenta accreta also incrementally increases, and is estimated at 3% at the first cesarean delivery followed by 11%, 40%, 61% and 67% following second, third, fourth, and fifth subsequent CS deliveries, respectively [10].

Imaging is the cornerstone of diagnosis for CSDs and PAS. Ultrasound (US) is the preferred first-line imaging modality for post-CS complications but may not provide adequate tissue resolution to characterize tissue defects due to limited field of view, patient body habitus or tissue quality, and operator experience, leading to potential for misdiagnosis. MRI may serve as a useful adjunct in the evaluation of post-CS uterine abnormalities because of its superior tissue contrast and large field of view [11]. MRI is oftentimes used when the US results are equivocal for PAS with high risk factors, or for preoperative planning in patients with confirmed PAS [12]. There is extensive evidence supporting the safe use of both modalities during pregnancy due to the absence of ionizing radiation. A systematic review including 23 studies noted an average ultrasound sensitivity of 90.72% and specificity of 96.94% for detecting PAS [13] versus sensitivity of 94.4% and specificity of 84.0% for MRI [13].

In this case report, we will discuss the imaging findings and subsequent management of a patient with a uterine window and suspected placenta accreta spectrum in the setting of a dichorionic, diamniotic twin gestation.

Case presentation

A 30-year-old gravida 9 para 3 patient was admitted to the antepartum service with a dichorionic, diamniotic twin pregnancy at 23 weeks gestation (dated by early first trimester sonogram) with abdominal cramping and pelvic pain that radiated to the lower extremities. Past obstetric history included 3 prior cesarean sections, prior uterine dehiscence in

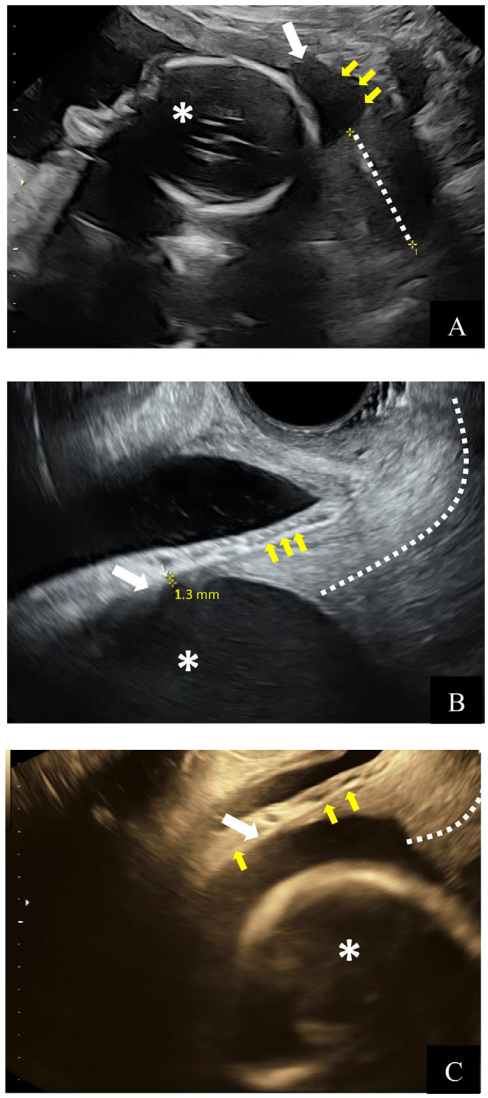

Fig. 1 – Transabdominal (A) 2D ultrasound of the presenting fetus at 23w6d demonstrating fetal head (asterisk), lower uterine segment bulge (yellow arrows), cervical canal (dotted white line), and location of myometrial thinning (white arrow). Transvaginal ultrasound (B) of the cervix, lower uterine segment, partially distended maternal bladder, and twin pregnancy at 22w6d. The white dotted line indicates the endocervical canal, the yellow arrows indicate the outer margin of the bladder serosa, and the white arrow indicates the thinnest portion of the lower uterine segment myometrium which measures 1.3 mm as indicated by the yellow caliber. Amniotic fluid without fetal presenting parts is visible (star). (C) Transvaginal ultrasound performed at 23w6d fetus with fetal head (asterisk) as the presenting part. Volume contrast imaging technique permits improved resolution of solid organ boundaries between the bladder wall and substantially thinned lower uterine segment myometrium. The yellow arrows indicate the distal margin of the maternal bladder, with the white arrow highlighting the proximal most region of nonvisualized myometrium. The dotted white line delineates the cervical canal.

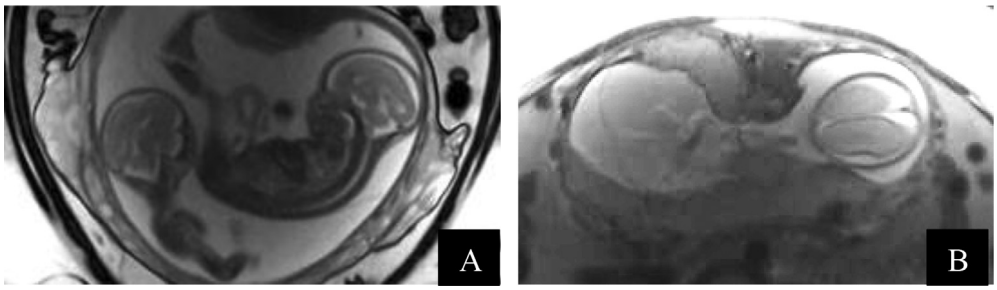

Fig. 2 – Coronal (A) steady state free precession and axial (B) single shot turbo spin echo T2 weighted images of the abdomen demonstrating a dichorionic, diamniotic twin gestation.

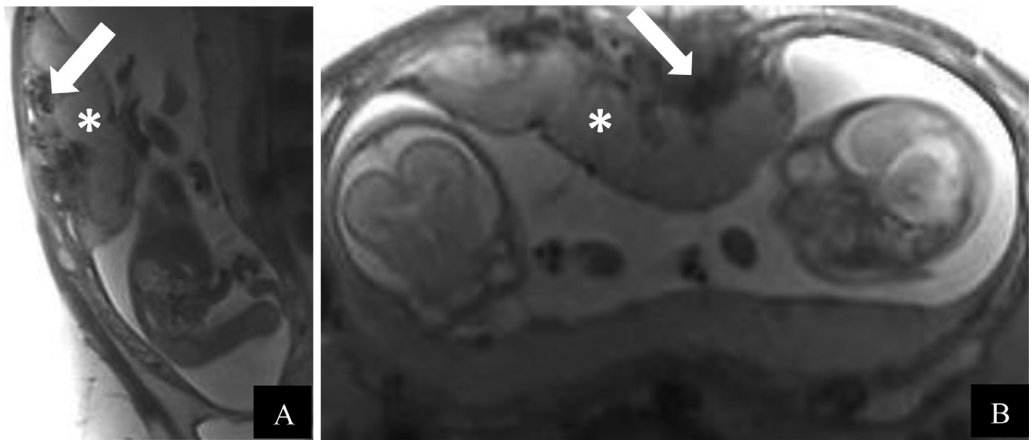

Fig. 3 – Sagittal (A) and axial (B) single shot turbo spin echo T2-weighted images of the abdomen demonstrating lobular and minimally heterogeneous anterior placenta (asterisk) with increased sub-placental flow voids (arrows).

her second C-section, and surgical site infection. Past medical history was relevant for iron deficiency anemia (hemoglobin of 7.8 mg/dL), gastroesophageal reflux disease, and obesity (BMI 53 kg/m^2). Upon admission she was found to have uterine contractions on tocometry without cervical change over serial examinations for preterm labor.

Prior to her admission, she underwent multiple ultrasounds, including routine 13- week ultrasound and 20-week anatomy scan, both of which demonstrated thinning of the myometrium in the anterior lower uterine segment to 1-2 mm (Fig. 1), concerning for uterine window. Ultrasounds also noted anterior and posterior placentas without placenta previa and without sonographic features of PAS. Ultrasound imaging was limited by acoustic shadowing, multiple gestations, maternal body habitus, and maternal abdominal scarring.

In order to further clarify these findings, the patient underwent abdominopelvic MRI at 22 weeks gestation that again showed a diamniotic, dichorionic twin gestation with fetuses in breech and transverse presentations (Fig. 2A). Respective placentas were located in the posterior and anterior uterine fundus and body (Fig. 2B). The posteriorly located placenta was normal in appearance. The anterior placenta was noted to be lobular with a focal area of increased subplacental vascularity concerning for PAS (Fig. 3) but without evidence of placenta previa. Focal myometrial thinning was noted in the anterior lower uterine segment, with a minimum myometrial thickness of 2 mm, as well as focal bulging of the amniotic sac with abutment of the bladder dome and perivesicular fat pad (Fig. 4).

Clinical management

Given the findings of suspected uterine “window” and concomitant focal PAS, the patient was deemed to be at increased risk of both uterine rupture in labor and severe peripartum hemorrhage. As her symptoms at the time of admission were mild, she was managed conservatively with Tylenol, heating pads, and abdominal binder as needed for pain. Daily fetal heart tracings were reactive and reassuring. Plans were made for repeat MRI at 28-30 weeks to re-evaluate the uterine window, and for a repeat growth US after one month. Delivery plan included repeat CS with midline vertical skin incision at 34 weeks. Due to personal issues requiring her departure from the hospital, she was discharged at 23 weeks 6 days. At the time of discharge, she was clinically stable with no pain, contractions, or leakage of amniotic fluid. The patient was counseled to return if she experienced symptoms such as pain, vaginal bleeding, contractions, fluid leakage, or decreased fetal movement. She continued with outpatient surveillance of

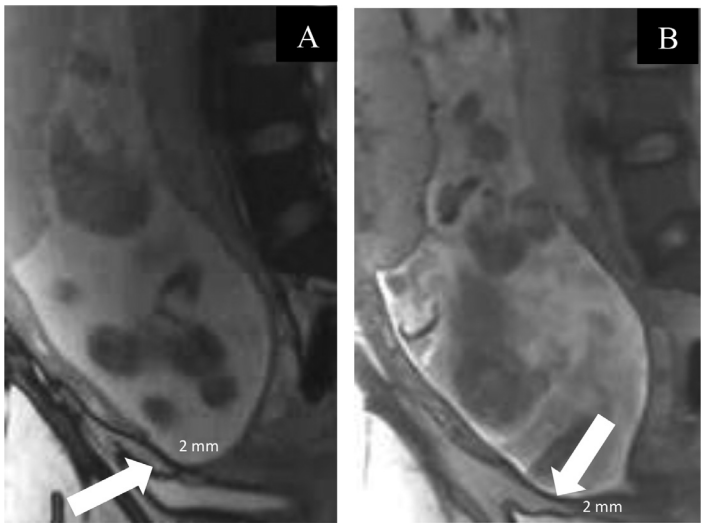

Fig. 4 – Sagittal steady state free precession (A) and single shot turbo spin echo (B) T2-weighted images of the abdomen and pelvis. Marked thinning of the myometrium is also noted in the lower uterine segment (white arrows) with 1-2 mm of myometrium remaining.

twin growth, placental appearance, and lower uterine segment appearance with serial ultrasound.

The patient returned to the hospital at 30w3d for routine monitoring. Her repeat ultrasound was less concerning for placenta accreta, and she did not undergo repeat MRI. Repeat ultrasound at 34w3d confirmed placental locations and appearances.

She underwent scheduled repeat cesarean section at 35w4d. She did not have contractions, vaginal bleeding, or fluid leakage at presentation and had adequate fetal movement. A midline vertical skin incision was performed to avoid significant adhesive disease expected in the lower pelvis based on surgical history and prior operative notes. The suspected uterine "window" was unable to be visualized intraoperatively due to obscuration by the urinary bladder, which overlayed it completely, and was not surgically dissected from the lower uterine segment. A mid-segment transverse hysterotomy was performed and both fetuses were delivered via breech position without nuchal cords and with normal Apgar scores. Both placentas were also delivered with gentle traction, and there was no intraoperative concern for PAS. Shortly after delivery, while recovering in the postanesthesia care unit, the patient developed postpartum hemorrhage unresponsive to uterotonic medications and requiring dilation and curettage. Ultrasound following onset of her symptoms demonstrated a thin endometrial stripe with an area of hyperechogenicity anteriorly, not contiguous with the endometrial cavity and concerning for stigmata of abnormal placental attachment.

The patient had significant dizziness in the days following, with decreased oral intake, consistent with anemia. Her preoperative hemoglobin was 11 mg/dL, with a postoperative hemoglobin of 7.3 mg/dL. Computed Tomography of the abdomen and pelvis performed on postoperative day 1 excluded active uterine bleeding or hematoma formation (Fig. 5). She was managed with 3 units of packed red blood

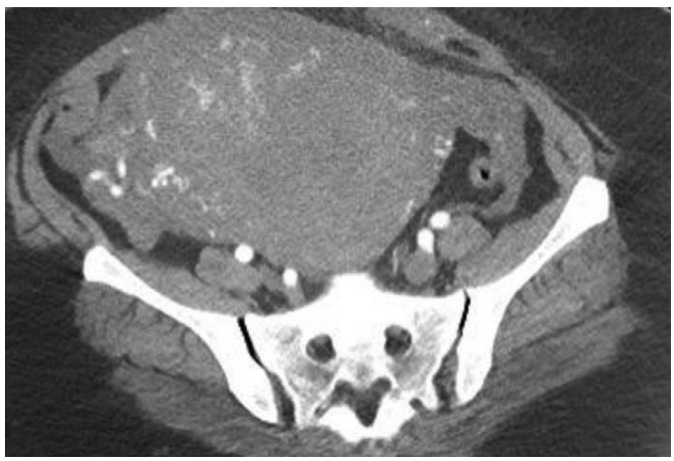

Fig. 5 – Axial CT of the abdomen with IV contrast demonstrating an appropriately enlarged and hypervascular postpartum uterus without evidence of active bleeding or intraabdominal hematoma.

cells and had an appropriate response, after which she was discharged.

Pathologic evaluation of both placentas revealed small size for gestational age, with villous dysmaturity, infarcts, focally increased intervillous/perivillous fibrin deposition, and calcifications. These microscopic findings may indicate placenta accreta spectrum, which is characterized by absence of villi and increased angiogenesis and protein deposition [14].

Discussion

We describe a case of cesarean scar compromise (uterine "window") by antenatal imaging with suspected concomitant placenta accreta spectrum in the setting of dichorionic diamniotic twin gestation. Both conditions may become increasingly common as rates of cesarean deliveries, ART and advanced maternal age increase.

Ultrasound is the first-line imaging modality for most abdominopelvic pathology in pregnancy. On ultrasound, large cesarean scar defects such as uterine window may appear as an area of myometrial thinning or cystic lesion in the anterior lower uterine segment, as was seen in this patient. Less severe lesions along the spectrum of CS scar compromise include uterine niche, which occurs in nonpregnant patients, which may appear as an anechoic triangular area in the region of the cesarean incision. When imaging findings of CSD are present on ultrasound, MRI may permit further characterization of the uterine wall and exclude other etiologies, such as adenomyosis, Nabothian cysts and leiomyomas or ultrasound artifact. MRI can also serve as a valuable tool for the purposes of surgical planning prior to repeat CS.

Cesarean scar compromise, or uterine window, is defined as asymptomatic complete separation of the myometrium with intact overlying serosa. This is usually clinically occult and incidentally noted at the time of Cesarean delivery or during routine ultrasound. The most feared complication of CS scar compromise is progression of this to uterine rupture, which involves complete disruption of all uterine layers (including serosa) with clinical signs of intraabdominal hemorrhage, and rarely occurs prior to labor. While there are not clear guidelines on the minimum myometrial thickness required to predict severe CS scar compromise, a systematic review that included 2776 patients reported that a myometrial thickness between 0.6 and 2.0 mm provided a strong positive predictive value for the presence of uterine dehiscence at delivery [15]. The classification of a cesarean scar defect has also been shown to be severe if the ratio between the myometrium at the scar level and adjacent myometrium is less than 50% [16]. When reporting uterine window and/or uterine dehiscence, it is also important to describe proximity of the defect to the urinary bladder and perivesicular fat pad, which may provide additional coverage and support of the thinned lower uterine segment prior to delivery.

Ultrasound is also the first line imaging modality for diagnosis of PAS. Features may include loss of the retroplacental clear space, myometrial thinning, anechoic placental lacunae, placental outpouching, and neovascularization [17]. MRI is indicated in isolated PAS with inconclusive ultrasound findings and/or risk factors for PAS. Detecting adjacent organ invasion, or further characterization of myometrial thinning can be aided through additional evaluation with MRI. Findings of PAS on MRI include T2 dark placental bands, lobular placental contour, placental bulge or mass, thinning or discontinuous myometrium, and in cases of placenta percreta, extension to or beyond the uterine serosa. In both ultrasound and MRI detection of PAS, sensitivity and specificity can vary by imaging unit, and can depend on quality of sonographic technique and equipment, as well as familiarity of radiologist who is trained to detect the subtle imaging findings.

Patients with PAS and a CSD have improved outcomes when managed with a planned cesarean section prior to the onset of labor, and hysterectomy with the placenta left in situ [18]. In this patient, repeat imaging was essential to understanding the persistence of the uterine window and determining the approach of operative delivery as well as delineating morbid placentation as the likely cause of her postpartum hemorrhage. Of note, repeat MRI was not performed which may have further delineated subtle features of PAS in the preoperative period.

Conclusion

As rates of cesarean deliveries are increasing, so too is the incidence of cesarean scar defects and placenta accreta spectrum. Both conditions demonstrate characteristic findings on ultrasound and MRI that may aid in diagnosis and influence subsequent clinical management and surgical planning. Timely diagnosis of both conditions is essential in order to decrease the changes of life-threatening hemorrhage and reduce maternal and fetal morbidity and mortality.

Patient consent

The patient reported in the manuscript signed the informed consent/authorization for participation in research which includes the permission to use data collected in future research projects including presented case details and images used in this manuscript.

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