

Case Report

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10.4103/pjog.pjog_11_21

A rare case of first-trimester placenta increta in an unscarred uterus: Diagnostic and management strategies

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Abstract:

Placenta accreta syndrome (PAS) is rare in first-trimester abortions with an unscarred uterus. It is this rarity that makes diagnosis and management difficult and challenging. This is a case report of a multigravid with an early incomplete abortion complicated by PAS (placenta increta) manifesting as an ill-defined hypervascular uterine cavity mass on transvaginal ultrasound, with decreasing trends of serum beta-human chorionic gonadotropin. PAS was successfully diagnosed preoperatively, and an uneventful hysterectomy was performed. A curettage that could potentially lead to catastrophic hemorrhage was prevented. This case highlights the diagnostic dilemma in early trimester PAS, the importance of early accurate diagnosis, and a good correlation with ancillary diagnostics to provide prompt and appropriate management.

Keywords:

Abortion, color Doppler, early pregnancy, placenta accreta, ultrasound

Introduction

Placenta accreta syndrome (PAS), formerly known as morbidly adherent placenta, is a potentially life-threatening cause of hemorrhage that contributes significantly to maternal morbidity and mortality. It is defined as an abnormal invasion of the placenta into the myometrium. PAS encompasses accreta, increta, and percreta, depending on the depth and extent of adherence. It is seen in 1 in every 2500–7000 pregnancies. PAS is often diagnosed in the third trimester, especially among women with cesarean section scar and placenta previa. However, first-trimester PAS diagnosed after an abortion is rare. Only 26 cases have been reported worldwide, four of whom had no previous uterine scar.^[1] Two cases were reported locally, both with scarred uteri.^[2,3] This case is the first in our institution, diagnosed preoperatively in a patient without any uterine scar,

confirmed histologically, and managed with hysterectomy with a good outcome.

The rarity of PAS recognized after a first-trimester abortion poses a diagnostic and management challenge because of the absence of established guidelines. An unsuspecting obstetrician who performs curettage for incomplete abortion puts the patient at risk for catastrophic hemorrhage. Management options other than hysterectomy are still evolving. This study will discuss the clinical presentation, the diagnostic and management strategies, and the importance of a high index of suspicion that will ensure prompt, individualized decisions needed for a successful outcome.

Case Report

A 39-year-old, Gravida 5 Para 3 (3013), married, Roman Catholic was admitted due to persistent vaginal spotting after a spontaneous abortion.

How to cite this article: Locsin SF, Tongco CB. A rare case of first-trimester placenta increta in an unscarred uterus: Diagnostic and management strategies. Philipp J Obstet Gynecol 2021;45:82-6.

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Submitted: 27-Apr-2021
Revised: 30-Apr-2021
Accepted: 28-May-2021
Published: 20-Jul-2021

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Her menses have been regular and monthly, with normal flow and duration. Her first three pregnancies were term, uncomplicated vaginal deliveries. Her fourth (2015) ended in a completed abortion with no curettage. Gynecologic, medical, family, personal, and social histories were unremarkable. Coitus interruptus was the only method of contraception.

Eight weeks before admission, the patient consulted a private physician for missed menses of 2 months and a 1-day episode of profuse vaginal bleeding with passage of meaty material. Pregnancy test was positive. The diagnosis was incomplete abortion at 8 weeks age of gestation. Completion curettage was advised but she refused for financial reasons. From then on, she experienced vaginal spotting which persisted such that she consulted at our emergency room 3 weeks before admission. Other than the spotting, she did not complain of dizziness, weakness nor abdominal pain. Physical examination showed pink palpebral conjunctiva, a soft abdomen with no palpable masses. On speculum examination, there were no lesions on the vaginal wall and cervix. On internal examination, the cervix was closed and smooth, uterus was not enlarged, nontender, and the adnexa were unremarkable. The diagnosis then was a completed abortion. Serum beta-human chorionic gonadotropin (B-HCG) and a transvaginal ultrasound were requested on outpatient basis.

On follow-up (2 weeks before admission), she reported cessation of the vaginal spotting. Physical examination findings were unremarkable. The initial serum B-HCG was elevated at 9532 mIU/ml. Transvaginal ultrasound showed "a normal-sized anteverted uterus measuring 5.53 cm × 6.58 cm × 5.07 cm; the endometrium was distinct and thin at the distal third of the uterine cavity [Figure 1]; there was an ill-defined mass measuring 3.67 cm × 2.30 cm × 2.48 cm extending from the middle to the upper third of the anterior endometrium [Figure 2], invading deep into the myometrium, with 0.26 cm of normal myometrium. Color Doppler revealed diffused dilatation of placental vessels surrounding the mass, with increased vascularity of the myometrial vasculature, highly suspicious of a gestational trophoblastic neoplasia (GTN) [Figure 3]. Complete blood count, coagulation studies, serum creatinine, and liver function tests were normal.

Serum B-HCG, repeated 7 days later, significantly decreased to 2700 mIU/ml. A repeat ultrasound showed similar findings as the first, except for a slight decrease in the size of the ill-defined mass to 3.36 cm × 2.92 cm × 2.85 cm. PAS (placenta increta) was considered and GTN was ruled out. The patient consented to a total abdominal hysterectomy with bilateral salpingectomy under spinal anesthesia.



Figure 1: Transvaginal ultrasound showing a normal-sized anteverted uterus measuring 5.53 cm × 6.58 cm × 5.07 cm. The endometrium is distinct and thin, measuring 0.4 cm at the distal third area

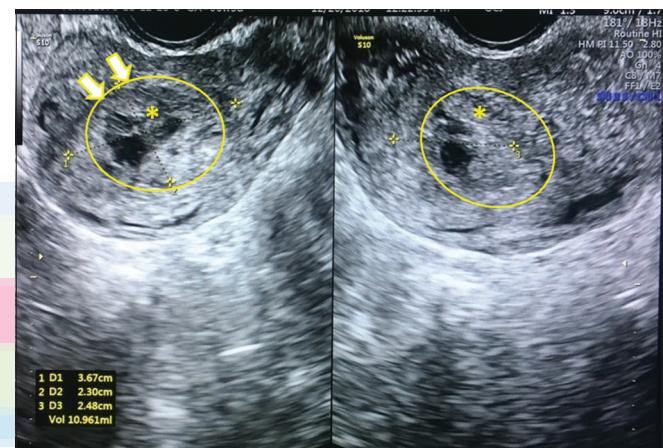


Figure 2: Transvaginal ultrasound showing an ill-defined mass (arrow) measuring 3.67 cm × 2.30 cm × 2.48 cm, extending from the middle to the upper third of the anterior endometrium, with thinning of the myometrium to 0.26 cm over the lesion and presence of lacunae (asterisk)



Figure 3: Color Doppler showing the ill-defined mass as hypervascular

On admission, physical examination findings were essentially unchanged from the previous evaluation

3 weeks ago. Intraoperatively, the uterus was slightly enlarged, measuring 7 cm x 5 cm x 4 cm with a smooth serosal surface. Ovaries and fallopian tubes were grossly normal [Figure 4]. On cut section of the uterus, there was a ragged, soft, friable, hemorrhagic mass within the uterine cavity at the right anterofundal area, measuring 2.7 cm x 2.7 cm and invading the myometrium, leaving 0.2 cm of the uterine wall intact. There was no gross adenomyotic lesion. The rest of the endometrium was smooth, measuring 0.2 cm [Figure 5]. Estimated blood loss was 250 ml. Postoperative course was unremarkable and she was discharged on the 3rd hospital day. One week postoperatively, serum B-HCG level remarkably decreased to 4 mIU/ml.

Histopathologic examination showed decidua, chorionic villi, and trophoblasts invading the myometrium, confirming the diagnosis of placenta increta. Endometrial glands and stroma were seen in the myometrium taken from sections overlying the mass, indicating adenomyosis [Figure 6a-d].

Case Report

With advances in imaging, PAS is often easily recognized in the third trimester of pregnancy, providing the obstetrician enough time to prepare strategies for a safe delivery. In the absence of an ultrasound diagnosis, PAS is recognized clinically when a difficult placental delivery is encountered and attempts at manual extraction end up in catastrophic bleeding, needing large volumes of blood transfusion, an emergency hysterectomy, and intra- and postoperative complications.

PAS that manifests as bleeding after a first-trimester abortion is rare and difficult to diagnose, as exemplified in our patient. She was diagnosed as incomplete abortion at 8 weeks age of gestation. Completion curettage at that time would have led to massive bleeding and possibly death. It is fortunate that she did not consent to this for financial reasons. If left unattended, the placenta could continue invading the myometrium and eventually rupture causing intraperitoneal hemorrhage. It is also rare to have PAS in an unscarred uterus, more so located on the fundal area. To our knowledge, she is the first case of a preoperatively diagnosed fundal PAS in an unscarred uterus in the first trimester with a successful outcome in our institution. Published reports from 3 authors (Timor-Tritsch, 2012; Shah, 2018; Wang 2019) show only 26 cases, but maternal survival outcomes were not included.

Our patient, with high parity and advanced age, had an initial clinical presentation of incomplete abortion. Even without curettage, her bleeding diminished but persisted for 5 weeks, which prompted consultation at our

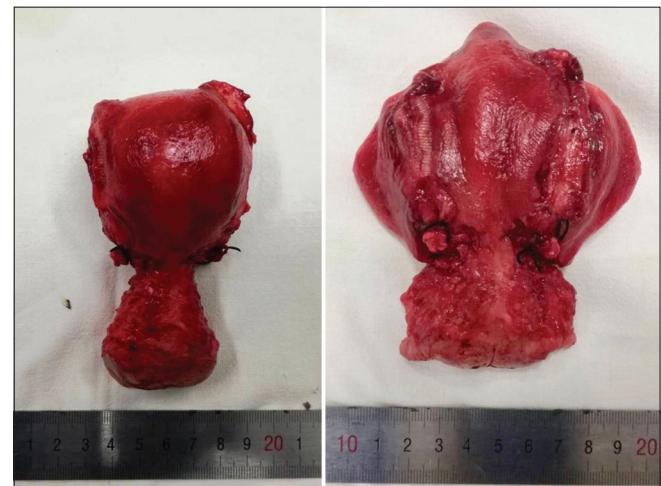


Figure 4: Gross specimen showing a slightly enlarged uterus measuring 7 cm x 5 cm x 4 cm with a smooth surface and regular contour

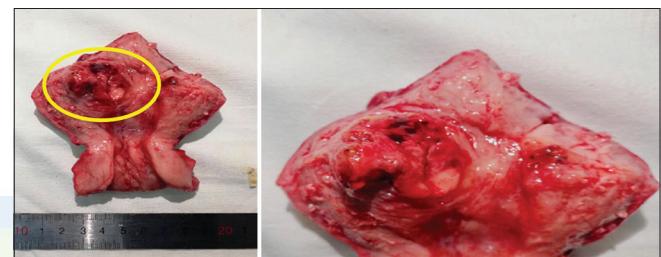


Figure 5: (Left) The gross specimen on cut section. (Right) There is a ragged, soft, friable intracavitory mass on the right anterofundal area, measuring 2.7 cm x 2.7 cm, with areas of hemorrhages, and invading into the myometrium, leaving 0.2 cm of normal myometrium intact. The rest of the endometrium is smooth, with 0.2 cm thickness

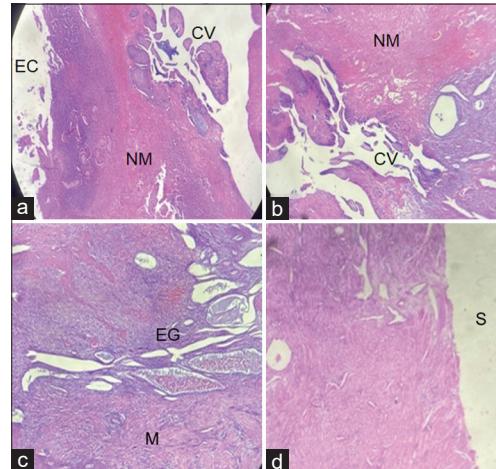


Figure 6: (a-d) Histopathologic examination (panoramic scanner view) showing placenta increta characterized by decidua, chorionic villi, and trophoblasts invading the myometrium. Endometrial glands and stroma were likewise seen in the myometrium indicative of adenomyosis. The endometrium has sheets of large polygonal cells with abundant cytoplasm. There are well-formed round to cystically dilated, tortuous glands with ballooning, cleared-out cytoplasm. There is no evidence of malignancy. (EC: Endometrial cavity, NM: Areas of necrosis in the myometrium, S: Serosa)

institution. Our baseline internal examination findings were unremarkable. Initial serum B-HCG level was high

and the ultrasound showed a vascular intracavitory mass on the fundus invading the myometrium. At this point, several conditions were considered PAS (increta), missed abortion, GTN probably invasive mole, placental site trophoblastic tumor (PSTT), epithelioid trophoblastic tumor (ETT), and uterine arteriovenous malformation (AVM). The correlation of clinical findings with ultrasound and color Doppler together with serial B-HCG monitoring helped clinch the diagnosis of PAS (placenta increta).

In a systematic review of first-trimester abortions with PAS by Wang *et al.*, the most frequent symptom is vaginal bleeding. Majority are massive after surgical abortion (65%), 35% have delayed onset (from 1 week to 2 years). Less than half have a complex mass in the uterine wall.^[1] Abdominal pain and rupture from the mass are late signs but catastrophic. Our patient presented with intermittent vaginal bleeding for 2 months and an ill-defined intracavitory uterine mass. She did not complain of abdominal pain or tenderness on examination even though there were sonologic signs of deep myometrial invasion.

What risk factors will raise our suspicion for first-trimester PAS? The strongest risk factor is a prior cesarean section (87%).^[1] Cesarean delivery was consistently seen in all three forms of PAS according to Klar. Some cases of cesarean scar pregnancy are now being considered as early PAS.^[4] However, there were also some reports of PAS in those without a uterine scar.^[5] Placenta previa, although significant in the second and third trimesters, is not seen in first-trimester scans so that this information is not helpful in early PAS. Other risk factors include those that damage endometrial integrity, such as curettage, manual placental removal, endometritis, myomectomy, hysteroscopic procedures, IVF, radiation, and even IUD insertion. Uterine pathology such as adenomyosis, submucous fibroids, and bicornuate uterus may also predispose to PAS. Advanced maternal age and multiparity are other risk factors. Advanced age even without a CS scar increases the odds of PAS by 1.3 for every year increase in age starting 35 years old. Failure to respond to multiple rounds of medical management for missed abortion should also prompt workup for PAS.^[5]

Several hypotheses have been proposed to explain the abnormal placentation in PAS. These include (1) abnormal trophoblast action, (2) a defect in the basalis layer of the endometrium from an iatrogenic mechanism, with abnormal healing and decidualization, and (3) abnormal vascularization and poor tissue oxygenation in the scar.^[6] That is why PAS is highly associated with a previous uterine scar (CS) or a history of endometrial trauma. It is very difficult to hypothesize how PAS developed in our patient. We were not able to

elicit any possible source of trauma to her endometrium. Adenomyosis was not seen grossly but was reported on histopathology from sections of the myometrium over the mass, but it was not extensive. Whether this provided the path to trophoblastic invasion on the uterine fundus is a big question. More recently, an entity called uterine body PAS has been described in 133 cases where PAS, which developed on the uterine body and fundus, eroded the myometrium and eventually lead to uterine perforation and intraperitoneal bleeding in many of the cases.^[7] Alternatively, could the retained products of conception from her recent abortion, which stayed on for 8 weeks and possibly became infected, be the trigger for abnormal trophoblastic invasion? In her condition, there seems to be more questions than answers.

The first-line diagnostic modality for diagnosis of PAS is a transvaginal ultrasound with color Doppler because of its high sensitivity (91%) and specificity (97%), aside from being cheap and easily available.^[8] Sonologic criteria in the third trimester have been clearly established in literature: placental lacunae, thinning of retroplacental myometrium, disruption of the echogenic interface between bladder and uterus with bridging vessels, and loss of the retroplacental sonolucent zone. Often, there is concomitant low-lying placenta or a previa. However, in the first trimester, after an abortion, the sonologic findings are nonspecific, with a sensitivity of 53.4%–74.4% and specificity of 70.8%–94.8%. These include a well-vascularized hyperechoic lesion, loss of endomyometrial interface, multiple lacunae, and myometrium thinning. In Wang's report, 43% of cases had a uterine cavity mass, majority of which were on the lower anterior wall.^[1] Color Doppler will show turbulent lacunar flow, increased subplacental vascularity, low resistance index, gaps in the myometrial blood flow, and vessels bridging the placenta to the uterine margin. All of these sonologic findings were seen in our patient. In viable first-trimester pregnancies with placenta accreta, a low implantation of the sac is the most common sign.

We did not request magnetic resonance imaging because of its cost. It has no additional advantage over ultrasonography, especially in the absence of placenta percreta and signs of local invasion.

Serum B-HCG monitoring was very helpful in our patient, in conjunction with ultrasound, in ruling out GTN. A steep decline is seen in completed abortion. When the decline is less than 60% after 7 days, retained trophoblasts are suspected. In our patient, there was a drop of 72%, which signifies an ongoing abortion rather than GTN. Her rapidly declining B-HCG and the vascular mass strongly suggested PAS. Remarkably, 1 week after our patient's hysterectomy, her B-HCG levels were almost nil (4 mIU/ml).

The other differential diagnosis was ruled out based on serum B-HCG trends and their ultrasound characteristics. Low serum B-HCG levels and absent or focal vascular flow suggesting the absence of invasion easily rules out missed abortion. Negative serum B-HCG and a hypervascular mass with flow reversal or high velocity, turbulent flow with low impedance are seen in AVM.^[9] Vascular uterine masses with persistently high HCG are seen in GTN, and low but nondecreasing trends are present in PSTT and ETT.

Many of the cases of first-trimester PAS who bleed after an abortion, reported before year 2012, were managed by abdominal hysterectomy. This reflects the urgent nature of the management as most of the patients presented with bleeding, rupture, and shock.^[4] More recently, laparoscopic hysterectomy in stable patients is gaining popularity. While this may be acceptable for patients who have completed their family size and do not wish to preserve childbearing potential, there are patients who are young and of low parity who would request uterine preservation. At present, medical and conservative surgical options have been reported with few successful outcomes. These options include cytotoxic therapy with methotrexate, uterine artery embolization alone or as a preoperative adjunct, transcatheter arterial chemoembolization using dactinomycin, hysteroscopic resection, laparoscopic hysterectomy with placental tissue removal, and high intensity focused ultrasound followed by hysteroscopic resection.^[10] Conservative management requires close follow-up as some cases bleed again, necessitating a hysterectomy. Future fertility remains to be elucidated. Patients should be made aware of another accreta in the next pregnancy, uterine AVM, uterine rupture, or ectopic pregnancy in the hysterotomy scar. All management options were presented to this patient but she preferred hysterectomy because she does not have future plans for pregnancy and she does not want to be burdened with cost of monitoring and compliance to regular follow-up. While the choice of management for her may be straightforward, the timing of hysterectomy could have been improved. In retrospect, a prompt hysterectomy should have been timed early on when a thinned-out myometrium was detected on her first ultrasound to avert the danger of uterine perforation by the invading mass. It would not have mattered whether her diagnosis was PAS, GTN or AVM. Hysterectomy is indicated in all these conditions, in a woman of completed family size, and most especially in the face of an impending emergency.

The importance of early recognition of this condition cannot be overemphasized. It gives the physician an opportunity to plan and organize a management team and avoid unexpected catastrophic bleeding. The challenge lies in identifying patients with rare PAS in

the first trimester who have no previous uterine scar or uterine manipulation, and with minimal symptoms, as in our case. It is, therefore, important to include PAS in the differential diagnosis of patients who persist to bleed after a first-trimester abortion.

Correlation of clinical presentation, serum B-HCG titers, and ultrasound will help rule out the many differentials. Prompt management, especially when signs of imminent perforation of the uterus are present on imaging, is necessary to achieve a successful outcome.

Declaration of patient consent

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

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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