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Case Report

Antepartum unscarred uterine rupture caused by placenta percreta: a case report and literature review

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Summary
The main risk for uterine rupture is the presence of a uterine scar due to prior cesarean delivery or other uterine surgery. However, rupture in an unscarred uterus is extremely rare, and risk factors include multiple gestations, trauma, congenital anomalies, use of uterotonics and placenta accreta spectrum.
Placenta accreta spectrum, also known as morbidly adherent placenta, is becoming increasingly common and is associated with significant maternal and neonatal morbidity and mortality.
We report a case of unscarred uterine rupture due to placenta percreta in a multiparous woman that required emergency peripartum hysterectomy.

Key words: placenta percreta, unscarred uterine rupture, hysterectomy, maternal morbidity

Introduction

Uterine rupture is a rare obstetrical condition that can result in life-threatening situations for both the mother and the fetus [1].
Although the main risk factor for uterine rupture is the presence of a uterine scar due to prior cesarean delivery or other uterine transmyometrial surgical incisions (myomectomy or adenomyomectomy), with a reported incidence of 23 to 81 per 100,000 deliveries [2], there have been reports of unscarred uterine rupture, occurring in 3.8 to 5.1 per 100,000 deliveries [3-4]. Risk factors for unscarred uterine rupture include multiple gestations, trauma, obstructed labor, congenital anomalies (such as Ehlers-Danlos type IV syndrome), use of uterotonics, and placenta accreta spectrum (PAS) [5-6].
Placenta accreta spectrum, formerly known as morbidly adherent placenta, refers to the range of pathologic adherence of the placenta, including placenta increta, placenta percreta, and placenta accrete [7].
In placenta accreta, the most common form, the villi adhere to the myometrium. Less commonly, the villi invade the myometrium (increta) or extend through the full thickness of the myometrium including the uterine serosa and sometimes adjacent pelvic organs (percreta) [8].
The most favored hypothesis regarding the etiology of PAS is that a defect of the endometrial-myometrial interface leads to a failure of normal decidualization in the area of a uterine scar from previous surgery (e.g., c-section) and only rarely in nulliparous without previous uterine surgery. This allows abnormally deep placental anchoring villi and trophoblast

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infiltration [9].

Antenatally, PAS can be diagnosed by obstetric ultrasound, especially if performed by experts, and by magnetic resonance imaging, even though in many cases it remains undiagnosed before delivery[9]. Although rare, uterine rupture due to PAS in a scarred uterus may occur due to the weakness of uterine layers caused by PAS in the second and third trimester [10,11]. The patient can present with vaginal bleeding, sudden or worsening abdominal pain, hemodynamic instability secondary to hemoperitoneum, and abnormal fetal heart rate tracing due to deterioration of fetal status [12].

On the other hand, very few cases of unscarred uterine rupture possibly caused by PAS have been reported [1,12-13].

We present a case of spontaneous uterine rupture in an unscarred uterus due to placenta percreta in a multiparous woman.

Case report

The patient was a G_5P_3 47-year-old woman with prior history of two uncomplicated pregnancies and two uterine cavity revisions (one for a spontaneous abortion and the other for therapeutic termination of pregnancy). She became pregnant by oocyte donation and intracytoplasmic sperm injection procedure. During pregnancy, the patient was diagnosed with gestational diabetes and treated with nutritional therapy.

At 39^{+0} weeks of gestation, the patient reported visual disturbances, headache and abdominal pain and was hospitalized for investigations, in the suspect of preeclampsia. Given the maternal age, gestational age, conception mode and gestational diabetes, induction of labor was performed with membrane stripping and oral misoprostol (25 mcg every two hours, with a maximum total dose of 200 mcg). Cardiotocographic monitoring was started at the beginning of induction and every four hours, as per hospital protocol.

After 12 hours, rupture of membranes and increasing uterine contractions were reported. The patient was sent to the delivery room for epidural analgesia, but which was not performed due to increasing patient discomfort, weak pulse, and paleness, as well as cardiotocographic evidence of fetal bradycardia. Regular uterine contractions and complete cervical dilation were documented.

A cesarean section was immediately performed due to abnormal fetal heart tracing. On laparotomy, a massive hemoperitoneum was observed and, shortly after the beginning of the surgery, the patient experienced a cardiac arrest, likely secondary to hemorrhagic shock. The patient was resuscitated and after a live male of 3600 gm was delivered and the placenta was manually removed, a wide tear through all layers of the right lateral and anterior uterine wall, extending from the fundus towards the paracervix, was noted. Given the extension of the uterine rupture and the hemodynamic instability, the decision was made to perform a total hysterectomy with bilateral salpingectomy. Total blood loss was 4200 ml.

Following surgery, the patient was transferred to intensive care unit for post-operative monitoring, and she recovered over the following weeks. At birth, the newborn had a funicular pH of 6.6, a SBE of -20.7 mmol/l and a 1, 5, and 10 min Apgar score of 3, 4 and 4, respectively. In addition, he was atonic, cyanotic, pale, unresponsive to stimuli, with a heart rate < 60 bpm, and was rapidly intubated with rapid normalization of the heart rate. Given the persistence of generalized hypotonia with absence of response to stimuli and lack of spontaneous respiratory activity, passive hypothermia was initiated. Cerebral function monitoring was performed, and the newborn presented a tonic-clonic seizure in the upper limb and widespread stiffness. After 7 days, the newborn was progressively extubated, maintaining stable vital parameters with nasal continuous positive airway pressure therapy. After another week, respiratory support was completely ceased. From a neurological point of view, he presented a hypoxic-ischemic brain damage due to uterine rupture. After 54 days of hospitalization, he was transferred to a long-term care facility.

The uterus, tubes and placenta were sent to the histopathology laboratory for analysis.

The main macroscopic findings for each specimen are reported below.

a Uterus of 23 x 13 x 14 cm with right parietal rupture of 10 cm.
b Right fallopian tube of 5 cm.
c Left fallopian tube of 4 cm.
d The placenta did not present any peculiar macroscopic findings.

Histopathological diagnosis

Each specimen described above was histologically evaluated and the diagnoses are reported below.

a Presence of villar structures penetrating through the myometrium and reaching the uterine serosa. These histological findings are compatible with placenta percreta.
b Adenomatoid tumor.

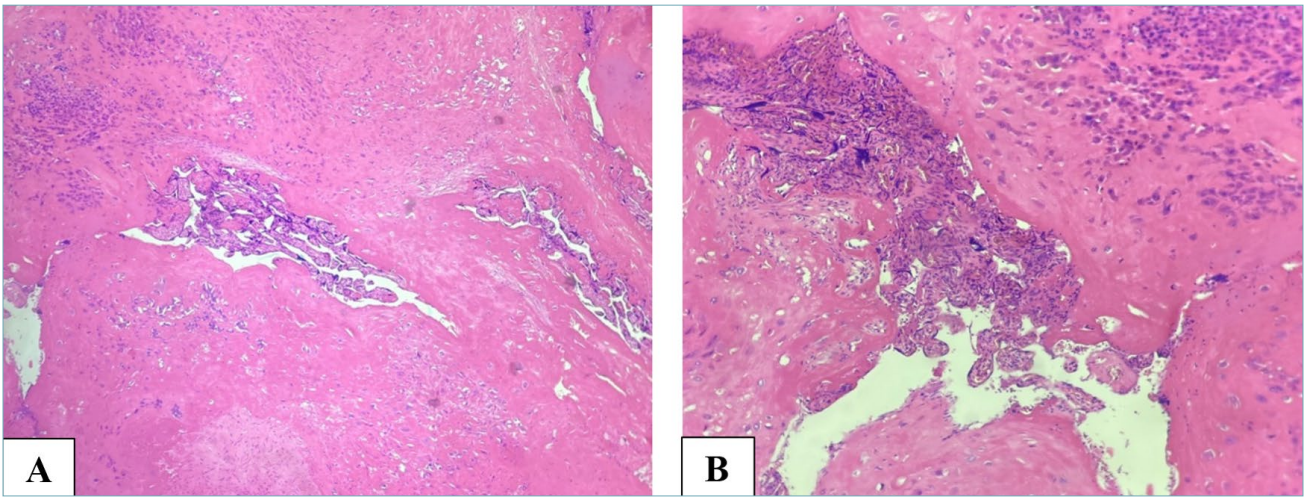

Figure 1. (A-B) Infiltration of the villar structures in the myometrium (A H&E 4x; B H&E 20x).

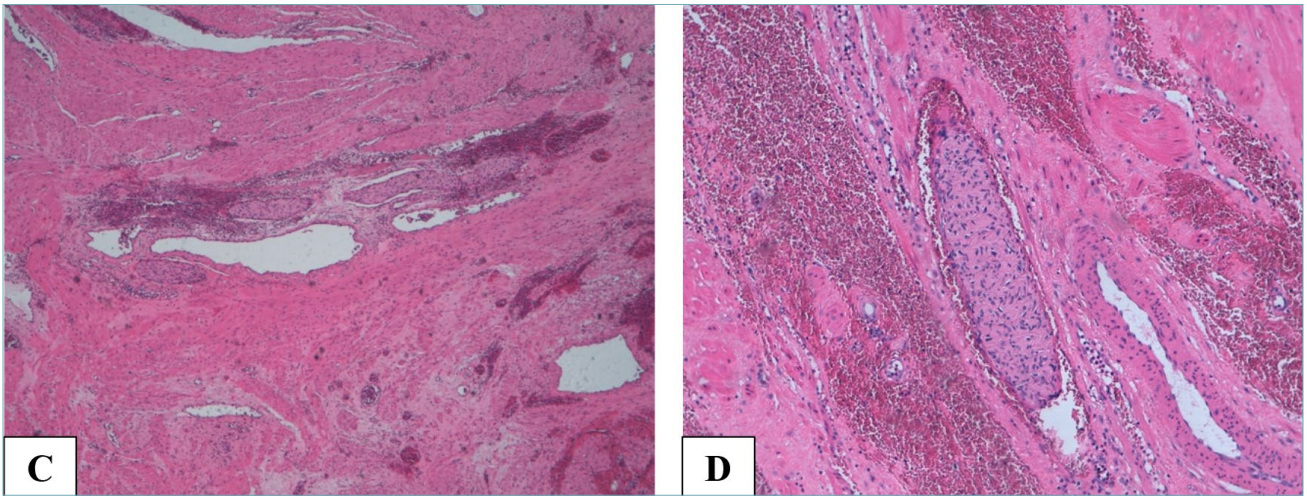

Figure 2. (C-D) Infiltration of the villar structures in the uterine serosa (C H&E 4x; D H&E 20x).

c No documentable neoplastic proliferation.

d The placenta shows a maturation of the chorionic villi compatible with the third trimester of pregnancy, with a prevalence of mature intermediate villi which appear crowded in different areas, mainly normoramified, with focal hyper-branching in the terminal villi.
An irregular subintimal thickening is observed at the level of the amniochorial vessels.
Presence of intervillous fibrin that configures fibrinoid pseudo-infarcts.

e Presence of funicle edema and minimal membrane inflammation.

Discussion

Complete uterine rupture is a rare peripartum complication that is often associated with high maternal and neonatal mortality and morbidity rates [6,15]. The main risk factor is the presence of a uterine scar, because of a previous cesarean delivery or uterine surgery, or following trauma [16]. Ruptures may occur in an unscarred uterus, although such cases are extremely rare. The goal of surgery is to control hemorrhage, identify injury of other intra-abdominal organs and minimize early post-surgical morbidity [15].
Of the identified risk factors for unscarred uterine rupture, placenta accreta spectrum, use of uterotonics,

Table I. Cases of uterine rupture due to placenta accreta spectrum.

Author (year)	Maternal Age (years)	Gravida/ Para	Gestational age (weeks)	Timing of uterine rupture	Histopathological diagnosis	Use of uterotonics
Okaniwa et al.(2021)[1]	29	G2P1	39 $^{+5}$	2 days postpartum	Accreta	Intravenous oxytocin, oral methylergometrine (after delivery)
Morken et al. (2001)[17]	25	G2P0	21	Antepartum	Percreta	-
Moldin et al. (1983)[18]	39	G4P3	33	Anterpartum	Percreta	-
Our case	47	G5P3	39	Antepartum	Percreta	Oral misoprostol

multiparity and advanced maternal age were present in our case [5]. Our patient was 47 years old and had two prior uterine curettages, which are known to increase the risk for PAS, but she did not have a history of prior cesarean section, which is the main risk factor for PAS. In this case, PAS was not suspected during routine prenatal ultrasound screening but was only diagnosed after histological evaluation of the placenta. With no suspicion of PAS, labor induction was performed with oral misoprostol – that, being a uterotonic, increased the risk of uterine rupture.

In the present case uterine rupture presented with some of the common clinical signs: maternal weak pulse and paleness, and cardiotocographic evidence of fetal distress. However, other signs such as change in uterine contractile activity, loss of fetal station or heavy vaginal bleeding were absent. Despite that, a high index of suspicion was still present and cesarean section was immediately performed.

Very few cases of unscarred uterine rupture possibly caused by placenta accreta spectrum have been reported in the literature (Tab. I) [1,17,18]. Of those reported, some occurred antepartum, like the one we described, and others postpartum.

Morken et al. [17] reported a case of a fundal uterine rupture at 21 weeks of gestation due to placenta percreta, which was diagnosed intraoperatively, in a young patient with a prior history of dilatation and curettage. Due to the young age of the patient and wish to maintain fertility, it was decided to resect only the uterine wall invaded and penetrated by the placenta. After 2 years, the patient was pregnant with a normal pregnancy delivered by cesarean section at 36 weeks [17].

Moldin et al. [18] reported a case of 2.5 cm fundal uterine rupture due to focal placenta percreta which was confirmed at the histologic examination. Given the limited extension of the pathological placental invasion in the uterine wall, it was decided to resect only the margins of the rupture and to suture the defect in the myometrium, instead of performing a hysterectomy. In that case, the patient had a history of prior uterine curettage complicated by uterine perforation, which may have contributed to the uterine rupture in placenta percreta [18].

More recently, Okaniwa et al. [1] reported a case of a 3 cm uterine rupture that occurred postpartum, as it was diagnosed two days after a vaginal delivery complicated by retained placenta, which was not manually removed. A total hysterectomy was performed and placenta accreta was confirmed by pathological examination. In PAS, postpartum uterine perforation or uterine rupture can be caused by manual removal of the abnormally adherent placenta [19,20], although in the case described by Okiniwa et al. manual removal of placenta was not attempted and placenta accreta likely was the main reason for unscarred uterine rupture [1]. In that case uterotonics had also been used.

Considering our case and the literature review, placenta accreta spectrum may be considered a risk factor for unscarred uterine rupture, which may occur either antepartum or postpartum. When PAS is suspected antenatally, even in an unscarred uterus, the rare possibility of uterine rupture must be kept in mind if suspicious clinical signs manifest.

Conflict of interest

The authors declare no conflicts of interest.

Ethics statement

Per Institutional policy, the review of medical records for publication of “case reports” of typically three or fewer patients is not considered human research and does not typically require IRB review and approval because case reporting on a small series of patients does not involve the formulation of a research hypothesis that is subsequently investigated prospectively and systematically.

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Author contributions

Conceptualization: EN, FC, VS; Data acquisition: IA; Writing: EN, VS, IA; Review: FC, MDT.

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