Amniocele associated with placental abruption: a case report

Bernard Gilles, MD; Fridjof G.S. Robergeau, MD; Larry M. Massena, MD; Maky-Kenson Nazaire, MD; Christophe Millien, MD, MMSc

Amniocele is a rare condition involving herniation of the amniotic sac through a uterine breach. Our case is of a 29-year-old pregnant woman at 31 weeks of pregnancy who presented to the maternity triage of the Mirebalais Teaching Hospital for abdominal pain and the passage of blood through the vagina. After an ultrasonographic evaluation, the diagnosis of amniocele was made. In practice, for a third-trimester, nonlaboring pregnant woman with this symptomatology, the most common diagnoses that come to mind are placenta previa and placental abruption. This case highlights that a diagnosis of silent uterine rupture should also be kept in mind knowing that a uterine rupture is a life-threatening event for both the mother and the fetus, therefore, early diagnosis is very important to improve the maternal-fetal prognosis.

Key words: 2D ultrasound, amniocele, Haiti, placental abruption, uterine rupture

Introduction

An amniocele is a herniation of the amniotic sac through a uterine defect and was first defined by Cotton et al in 1982.[1] To date, only about 30 cases of amniocele have been described in the literature.[2] It can occur through an undiagnosed uterine rupture. The latter is the occurrence of a breach in the wall of the uterus. Complete rupture involves rupture of all layers of the uterine wall, leading to spillage of uterine contents into the abdominal cavity, whereas an incomplete rupture has an intact peritoneum or serosa.[3]

More than 90% of the cases of uterine rupture occur in women who have already undergone a cesarean delivery during labor.[4] Silent ruptures have also been reported after a dilation and curettage and hysteroscopic procedures. The difficulty in the diagnosis and management thereof arises in the event of chronic and silent uterine rupture.[5] Amniocele through a silent uterine rupture can also be associated with placental abruption.[6] Uterine rupture is a catastrophic obstetrical complication that is associated with high rates of perinatal and maternal morbidity and mortality.[7]

We report a rare case of amniocele discovered fortuitously during triage at the Mirebalais Teaching Hospital with a good maternal-fetal outcome.

Case Report

This was a case of a 29-year-old, G3P1A1Lc2 patient who was seen in the maternity triage of the Mirebalais Teaching Hospital for hypogastric pain with light vaginal bleeding. The patient traced her symptomatology to about 4 days before the consultation. The patient had no particular medical or family history. In terms of her obstetrical history, her first pregnancy ended in abortion through dilation and curettage; for her second pregnancy, she underwent a cesarean delivery about 4 years previously for a breech or cephalic twin pregnancy. We were unable to obtain details of the surgery performed. The current pregnancy was followed irregularly because of a lack of economic means from the fifth month and ended at 31 weeks + 4 days gestational age according to the date of her last menstrual period at the time of the evaluation. No toxic habits were reported.

Upon physical examination, her vital signs were within the normal limits and the cardiopulmonary evaluation was unremarkable. She had an enlarged abdomen with an irregular outline, lower right palpation of a firm mass similar to a gravid uterus, upper left palpation of a large renitent mass (Figure 1), and the presence of a transverse suprapubic scar. There was no evidence of uterine contraction. The vaginal examination was unremarkable.

An ultrasound showed a single intrauterine fetus with cardiac activity in breech presentation with anhydramnios associated with a large anechoic mass that was initially mistaken for a giant ovarian cyst. However, after a second reassessment, the anechoic mass turned out to be the protruding amniotic sac containing the fetal left forearm and part of the umbilical cord through an approximately 5 cm antero-fundal uterine rupture (Figure 2, A). The placenta was also inserted anterofundically with lifting of the placental margin and a retroplacental hematoma at its distal portion in contact with the point of uterine rupture (Figure 2, B); there was no evidence of fluid effusion in the peritoneal

From the Department of Obstetrics and Gynecology, Mirebalais Teaching Hospital, Mirebalais, Center, Haiti

The authors report that they have no known competing financial interests or personal relationships that could have seemed to influence the work reported in this article.

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Corresponding author: Bernard Gilles, MD.
gillesbernard91@gmail.com

2666-5778/$36.00

http://dx.doi.org/10.1016/j.xagr.2023.100270

FIGURE 1
Enlarged abdomen with irregular outline of gravid uterus and amniocele

Gilles. Amniocele with placental abruption. Am J Obstet Gynecol Glob Rep 2023.

cavity. Cardiotocographic evaluation was unremarkable. The patient's hemoglobin level was 10.3 g/dL, her white blood cell count was 8100/μL, and the platelet count was 265,000/μL. The diagnosis of amniocele complicated by placental abruption on a silent rupture of a scarred uterus with an ongoing pregnancy of 31 weeks + 4 days was made. Consequently, she was urgently prepared for the operating room.

A midline sub umbilical incision was made up to the entrance to the abdominal cavity (Figure 3, A). After rupture of the amniocele (Figure 3, B), a classic hysterotomy in continuity with the point of rupture was performed to extract a male fetus weighing 1.65 kg with Apgar scores of 7, 8, and 9 at 1, 5, and 10 minutes of life, respectively (Figure 3, C). After resection of the old edges of the amniocele area, uterine closure was performed in 2 planes (Figure 4, A–B). The remainder of the procedure was uneventful with blood loss within the normal limits. The newborn was transferred to the neonatal intensive care unit for prevention and management of possible complications related to prematurity. After advice on her postoperative follow-up and choice of a family planning method (implant), the mother was discharged 5 days postoperatively without complications. Reviewed 4 months later at a follow-up, the mother and her child were in very good health.

Discussion

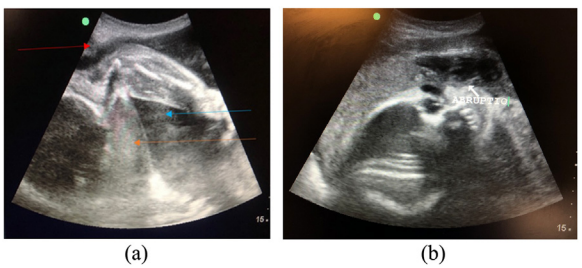
The presence of a uterine breach or weakness in the uterine wall is most often secondary to poor uterine healing. This may be the consequence of a previous uterine surgery, such as a myomectomy or cesarean delivery, or a complication of previous instrumentation of the uterus, such as dilation and curettage. Our patient presented with at least 2 of these risk factors. Rarely, it may be an incidental finding in an unscarred uterus.[8]

In cases of amniocele, the fundal localization is the most frequently found (58%), as was the case in our patient, and can be linked to a history of curettage, myomectomy, or uterine rupture. Localization at the level of the lower segment of the uterus is the second most common location (39%) and may be related to a previous cesarean delivery.[2]

In the literature, discovery of an amniocele was possible with a routine ultrasound examination in 29% of cases, but in most cases (71%), it was symptomatic with abdominal pain as the main symptom (61%), followed by a feeling of a uterine cramp (12.9%) or vaginal bleeding (9.7%).[2] Two cases of uterine rupture following placental abruption with massive blood effusion into the myometrium causing weakness at the level of the latter have been listed in the literature.[8] Our patient presented to triage with abdominal pain accompanied by light vaginal bleeding, and the fetus showed no signs of distress despite the apparent anhydramnios inside the uterine cavity. The patient was hemodynamically stable. Initially, we mistook the amniocele for a giant ovarian cyst, which was also the case in the studies by Casather[9] and Wali.[10]

Because antepartum uterine rupture is a life-threatening event for both the mother and the fetus, prompt and accurate diagnosis is very important to improve prognosis. The general belief is that a herniated amniotic sac in the abdominal cavity is strongly suggestive of an impending uterine rupture.[11] However, uterine rupture does not always occur immediately after ultrasound detection of an amniotic sac protrusion because a thin layer of myometrium and even uterine serosa may be around the herniated sac, which can be difficult to detect on ultrasound.[12] Magnetic resonance imaging

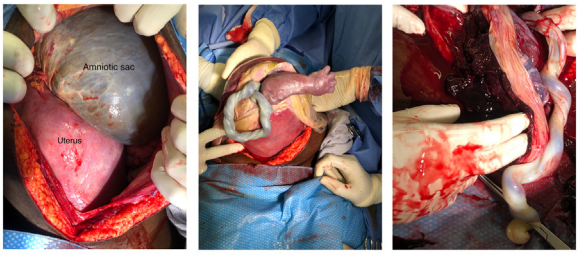
FIGURE 2
Pre-operative ultrasound images A, **Uterine rupture with protrusion of the fetal forearm.** ***Red arrow,*** **Proximal portion of the uterine muscle.** ***Orange arrow,*** **Distal portion of the uterine muscle.** ***Blue arrow,*** **Amniotic fluid.** B, **Visualization of placental abruption.**



(a) (b)

Gilles. Amniocele with placental abruption. Am J Obstet Gynecol Glob Rep 2023.

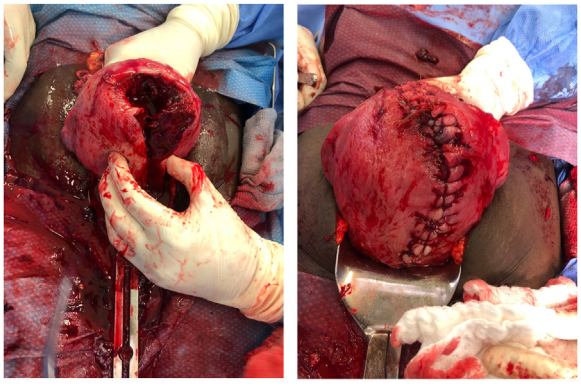
FIGURE 3
Intraoperative images A, **Uterine rupture with protrusion of the amniotic sac.** B, **Protrusion of the fetal left forearm and umbilical cord through the ruptured uterus.** C, **Visualization of the retroplacental hematoma.**



(a) (b) (cb)

Gilles. Amniocele with placental abruption. Am J Obstet Gynecol Glob Rep 2023.

FIGURE 4
Intraoperative images A, **Uterus after hysterotomy and delivery.** B, **Uterine closure.**



(a) (b)

Gilles. Amniocele with placental abruption. Am J Obstet Gynecol Glob Rep 2023.

(MRI) is necessary to assess this myometrial layer and its thickness.[10] MRI has unique advantages in the differential workup of the acute abdomen in hemodynamically stable pregnant patients and has demonstrated superior accuracy in measuring uterine wall breaches and abnormalities.[13] Obtaining an MRI in our hospital was not possible because it was not available.

The management of an amniocele depends on several factors including the desire to continue the pregnancy, the gestational age at the time of diagnosis, the fetal viability, the severity of the symptoms, and the size of the uterine breach. Before fetal lung maturity, 2 management protocols can be considered; these are surgical management and conservative management.[12] Surgical intervention involves termination of the pregnancy and repair of the uterine wall defect, whereas conservative management involves close monitoring. Most cases in which prolongation of the pregnancy was attempted ended up with deliveriy within a few hours or days after the diagnosis. This greatly shifts the balance of risks and benefits; conservative management usually has minimal benefit in prolonging pregnancy and includes risks of stillbirth and fetal asphyxiation; prompt delivery mitigates these risks. If patients elect conservative management despite counseling on the pros and cons, at the very least, antenatal corticosteroids should be considered, and the patient should be admitted for observation in a facility where immediate cesarean delivery facilities and neonatal intensive care are continuously available. The decision to repair the defect or perform a hysterectomy is based on a combination of factors including the extent of uterine damage caused by the rupture, the patient's desire for a future pregnancy, their intraoperative anesthetic and hemodynamic stability, and the surgeon's experience in repairing a complicated rupture.[6] To date, there is no consensus on the therapeutic strategy to adopt when faced with discovery of an amniocele.[2] Given our context, we immediately opted for termination of the pregnancy.

Uterine rupture is a life-threatening emergency that leads to maternal death in about 1% of developed countries and in 5% to 10% of developing countries.[14] The reported perinatal mortality rate associated with uterine rupture ranges from 5% to 26%.[6] Fetal death occurs more frequently in cases of placental abruption and/or fetal protrusion into the abdominal cavity.[6] Very few cases of silent uterine rupture with delivery of a healthy fetus have been reported so far,[15,16] and we can say that because of the expertise and vigilance of the care team, our patient and her newborn did well. Because of a lack of financial means, the patient was unable to make her prenatal visits early and regularly, which prevented her from having her amniocele diagnosed beforehand and could have compromised her prognosis in the face of this type of complication.

Conclusion

An amniocele is a rare event, and its association with a placental abruption, as in this case, is even rarer. A high index of suspicion for uterine rupture should be considered if there is a previous scar or procedures on the uterus, when there are unusual clinical features, and suspicious ultrasound findings, such as bands, cysts, or unexplained oligohydramnios. Ultrasonography has an important role in the early diagnosis of silent uterine ruptures, especially in resource-limited locations where MRI is not available. Especially because uterine rupture is a potentially fatal event for the mother and the fetus, early diagnosis is very important to improve the maternal-fetal outcome. The development of a health system with adequate materials and equipment that promotes

social protection of pregnant women is necessary to facilitate access to care for this population to allow early diagnosis in a low-resource environment. ■

REFERENCES

1. Cotton DB. Infant survival with prolonged uterine rupture. Am J Obstet Gynecol 1982;142:1059–60.

2. Azzaoui JE, Chmichi N, Slaoui A, et al. A large amniocele with protruded right arm and hemithorax: a case report and review of the literature. World J Pharm Life Sci 2019;5:28–36.

3. Sunanda N, Ranganth P. A two-year analysis of uterine rupture in pregnancy. Int J Reprod Contracept Obstet Gynecol 2016;5:3983–6. https://doi.org/10.18203/2320-1770.ijrcog20163875.

4. Miller DA, Goodwin TM, Gherman RB, Paul RH. Intrapartum rupture of the unscarred uterus. Obstet Gynecol 1997;89:671–3.

5. Conturso R, Redaelli L, Pasini A, Tenore A. Spontaneous uterine rupture with amniotic sac protrusion at 28 weeks subsequent to previous hysteroscopic metroplasty. Eur J Obstet Gynecol Reprod Biol 2003;107:98–100.

6. Yang SW, Yoon SH, Yuk JS, Chun KC, Jeong MJ, Kim M. Rupture-mediated large uterine defect at 30th gestational week with protruded amniotic sac and fetal head without fetal compromise after laparoscopic electromyolysis: case report and literature review. Medicine (Baltimore) 2022;101:e32221.

7. Hofmeyr GJ, Say L, Gülmezoglu AM. WHO systematic review of maternal mortality and morbidity: the prevalence of uterine rupture. BJOG 2005;112:1221–8.

8. Aiyekomogbon JO, Ojah SO, Shinkafi SM, Agom B. A large amniocele through a fundal uterine defect diagnosed on 2D ultrasound imaging. Int J Case Rep Images 2017;8:776–81.

9. Casather DM. VP55.01: Pregnancy with an amniocele, conservatively managed until fetal maturity: a case report. Ultrasound in Obstet & Gynec 2020;56:306.

10. Wali AS, Naru TY. Silent uterine rupture of scarred uterus—an unusual presentation as amniocele. J Ayub Med Coll Abbottabad 2013;25:204–5.

11. Oyelese Y, Tchabo JG, Chapin B, Nair A, Hanson P, McLaren R. Conservative management of uterine rupture diagnosed prenatally on the basis of sonography. J Ultrasound Med 2003;22:977–80.

12. Iemura A, Kondoh E, Kawasaki K, et al. Expectant management of a herniated amniotic sac presenting as silent uterine rupture: a case report and literature review. J Matern Fetal Neonatal Med 2015;28:106–12.

13. Ponder KL, Won R, Clymer L. Uterine rupture on MRI presenting as nonspecific abdominal pain in a primigravid patient with 28-week twins resulting in normal neurodevelopmental outcomes at age two. Case Rep Obstet Gynecol 2019;2019:2890104.

14. Gerard G, Nahum M. FACS. Uterine rupture in pregnancy.Medscape; 2021. Available at: https://reference.medscape.com/article/275854-overview. Accessed June 23th, 2023.

15. Rana R, Puri M. Pre-labor silent rupture of unscarred uterus at 32 weeks with intact amniotic sac extrusion: a case report. Cases J 2009;2:7095.

16. Woo JY, Tate L, Roth S, Eke AC. Silent spontaneous uterine rupture at 36 weeks of gestation. Case Rep. Obstet Gynecol 2015;2015:596826.