Case Report

Delayed stillbirth by hysterectomy following early-term uterine rupture with fetal demise in secundigravida

Shanshan Wang, PhD,a Muhetaerjiang Kadeer, MD,a Rouzi Yusufu, MMa, Junqiao Niu, MMA, Yan Liu, MD,a Patiman Rouzi, MD,b Shuang Sui, PhDc, Jia Wang, MMa, Xiaojuan Li, MMA, Yan Wang, MDa,b, Yongfang Ren, MMa,b,c,∗ Ying Huang, MMa,c,∗

aDepartment of Radiology in People’s Hospital of Xinjiang Uygur Autonomous Region
bDepartment of Obstetrics and Gynecology in hospital of Urumqi Friendship hospital, Xinjiang Uygur Autonomous Region
cDepartment of Obstetrics in hospital of Xinjiang Uygur Autonomous Region

A R T I C L E  I N F O
Article history:
Received 20 February 2021
Revised 30 April 2021
Accepted 30 April 2021

Keywords:
Uterine rupture
Postterm pregnancy
Fetal demise
Amniotic sac
Cornual pregnancy
Cesarean section scar

A B S T R A C T
Uterine rupture and postterm pregnancy pose a number of life-threatening complications to both mother and child, including severe intra-abdominal bleeding and peritonitis, birth injury, hypoxia, and fetal loss. This report presents a rare case of a 20-year-old female experiencing fetal demise at 60 weeks of pregnancy, with uterine rupture and bone tissue discharge from her vagina without severe intra-abdominal bleeding and peritonitis. The mild clinical course despite complete uterine rupture was due to the firm adhesion of the amniotic sac to the uterus caused by inflammation. The adhesion of the intestines to the rupture site prevented dehiscence of the ruptured wound. Suppression and bone tissue discharge relieved the pressure on the patient’s abdominal cavity and prevented subsequent occurrence of severe peritonitis. Radiologists mistakenly regarded the thick amniotic sac wall on the right side of the uterine wall as a right cornual pregnancy with uterine rupture caused by chronic inflammation. This report aims to bring awareness of this rare condition to medical students and radiologists.

© 2021 Published by Elsevier Inc. on behalf of University of Washington.
This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/)

Abbreviations: AMA, Advanced maternal age; CT, Computed tomography; MRI, Magnetic Resonance Imaging.
∗ Competing Interest: There is no conflict of interest
∗ Corresponding author.
E-mail addresses: wangyandoct@sina.com (Y. Wang), 2630942966@qq.com (Y. Ren), 3041791194@qq.com (Y. Huang).
https://doi.org/10.1016/j.radcr.2021.04.085
1930-0433/© 2021 Published by Elsevier Inc. on behalf of University of Washington. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).
Introduction

Uterine rupture is a rare and life-threatening complication for both mother and child [1,2]. It usually occurs in scarred uterus with intra-abdominal hemorrhage and peritonitis [3,4]. Postterm pregnancy also poses a number of risks to the fetus, including meconium aspiration, birth injury, hypoxia, and death [5-7]. This report presents a rare case of uterine rupture with fetal demise in a patient with mild symptoms at 60 weeks of gestation and explores the cause of its occurrence. To the best of our knowledge, such a case has not been reported before.

Case report

A 20-year-old female reported to People's Hospital of Xinjiang Autonomous Region in the 60th week of her undelivered pregnancy with low fever, pus, and bone tissue discharge from her vagina for further treatment (Fig. 1). Patient vital signs were as follows: pulse, 126 bpm; blood pressure, 92/64 mmHg; temperature, 38.6°C; and respiratory rate, 20 breaths/min. Gynecological examination showed that the patient's vagina was unobstructed, with purulent secretions and odor present. A fetal bone was located at the orifice of the cervix (Fig. 2).

Between 36th and 38th gestational week, the patient experienced severe abdominal pain, vaginal bleeding, suppuration, low fever, and then bone tissue discharge from her vagina (Fig. 1). The ultrasound examination at a local hospital demonstrated uterine rupture, fetal demise, and pelvic empyema. The patient received treatment at a local hospital, and the aforementioned symptoms were alleviated except for the bone tissue discharge from her vagina (details are unknown).

The patient's first child was born 4 years ago through a cesarean section. She had no relevant family history and did not drink alcohol or smoke.

Preoperative magnetic resonance imaging in our hospital revealed an intrauterine pregnancy. The uterus was enlarged, especially at the right uterine horn. Breech presentation of the fetus was observed, with a fetal arm protruding from the uterus into the abdominal cavity (Fig. 3). Ascites was also present. Preoperative computed tomography (CT)-further revealed that there was a considerable amount of gas in the uterus, the fetal bones below the two knees were missing, a fetal hand protruded into the patient's abdominal cavity, and a fetal femur bone protruded towards her vagina (Fig. 4,5). The patient's condition was diagnosed as right cornual pregnancy with uterine rupture that had resulted in fetal demise.

Since the severity of pelvic infection, uterine rupture, and uncertain anatomical relationship between the uterus, aorta, and ureters, massive hemorrhage during exploratory laparotomy could have been life-threatening. Preserving the uterus was more likely to cause continuous infection or other

Fig 1 – Bone tissue discharged from the patient's vagina at the local hospital.

Fig 2 – Gynecological examination showed a fetal bone at the orifice of the cervix (white arrow).

Fig 3 – Coronal MRI showed the fetus was in breech position, and uterus was enlarged, especially at the right uterine horn (white arrow). Uterine wall is shown with green arrow. Fetal hand protruded into patient abdominal cavity (yellow arrow), and fetal femur bone protruded into the vagina (blue arrow). (Color version of figure is available online)
complications. Imaging examination revealed that a fetal arm punctured the uterine wall. Thus, a surgical operation could damage the intestinal tract and the ureter. Therefore, exploratory laparotomy, abdominal aortic sacculus implantation, and bilateral ureteral stent implantation were performed.

During the operation, bilateral ureteral stent implantation through the bladder was first performed by the urologists. During exploratory laparotomy, a large mass closely adhering to the abdominal wall, omentum, small bowel, and retroperitoneum was found in the patient’s abdominal and pelvic cavity, with pus and odor and present (Fig. 6). Gastrointestinal surgeons successfully separated the adhesion between the mass, intestine, and retroperitoneum. Then, obstetricians explored the pelvic cavity. There was an extensive rupture at the cesarean section scar in the anterior wall of the patient’s uterus.

The deceased fetus was 20 cm long, with an incomplete amniotic sac protruding from the uterine scar into the abdominal and pelvic cavities. The amniotic sac was 0.5 cm thick and formed a thick cystic wall surrounding the fetus (Fig. 6). The fetal head was confirmed to be infected with Staphylococcus epidermidis via pus culture. The necrotic right hand protruded out of the amniotic sac, and the tissue below the two knee joints was lost (Fig. 7). The patient’s ovaries and fallopian tubes were severely swollen and deformed. As a result, hys-
terectomy and bilateral salpingectomy were performed, while patient's highly edematous ovaries were preserved. Pathological examination of tissue samples from the uterus, bilateral fallopian tubes, placenta, and fetal membrane showed massive degenerative villi and tissue calcification attached to the inner wall of the uterus. This structure's anatomical relationship with the myometrium was unclear. Focal abscess formed in the myometrium and serosa, and granulation tissue proliferated. There was diffuse infiltration of lymphocytes and plasma cells in the stroma of bilateral tubal mucosa (Fig. 10). The pathological examination results were consistent with what was found during the operation.

After returning to the ICU, the patient's body temperature returned to normal on the second day. The patient was discharged from the hospital 17 days after the operation.

Discussion

This report presents a rare case of uterine rupture and fetal demise in a young female with mild symptoms of intra-abdominal bleeding and peritonitis. Once the uterus ruptures, the flow of amniotic fluid and blood is expected to cause severe abdominal pain and acute peritonitis [8-10]. We hypothesize that the amniotic sac did not break following the uterine rupture, and firm adhesion to the uterine wall caused by inflammation prevented the dehiscence of the rupture wound, thus reducing blood flow in the uterine cavity and resulting in this specific clinical course. The partially broken amniotic sac caused by chronic inflammation was 0.5 cm thick as confirmed by pathological examination, which is an extremely rare condition. This is why the radiologists mistakenly regarded the thick amniotic sac wall around the fetus on the right side of the uterus as a right cornual pregnancy with uterine rupture (Fig. 9).

CT results further revealed a considerable amount of gas in the uterus. We speculate that the reasons for gas accumulation around the fetus are: one. bacterial infection, which was confirmed as Staphylococcus epidermidis via bacterial pus culture; and 2. bone tissue discharge from the patient’s vagina, which formed a channel for gas to enter the patient’s uterus.

During the operation, obstetricians and gynecologists found an extensive rupture at the cesarean section scar in the anterior wall of the patient's uterus. Her ovaries and fallopian tubes were severely swollen and deformed. They suggested that the patient’s uterus had been ruptured for at least 20 weeks, and her uterus and bilateral fallopian tubes were seriously infected. Therefore, obstetricians and gynecologists decided to remove her uterus and bilateral fallopian tubes and preserve the ovaries.

In the most recent literature review of cases involving uterine rupture, the majority of uterine ruptures were found to occur in women with a history of uterine surgeries [11]. The case described here is unusual because the patient did not present with typical clinical symptoms. It is possible that the
The patient previously recovered. Diabetes, placental abruption, and abnormal placental locations had increased rates of hypertensive disorders, diabetes, placental abruption, and abnormal placental locations. Young mothers have less concomitant pregnancy- and delivery-related risk factors associated with adverse neonatal outcomes. The mother’s young age in this case also played an important role in the mild clinical course and fast recovery.

One of the limitations of this report is that the patient had previously received treatment at a local hospital, and we were unable to retrieve the medical and radiographic records of the patient from that hospital.

### Patient consent:

The patient has provided informed consent for publication of the case.

### References

[1] Dow M, Wax JR, Pinette MG, Blackstone J, Cartin A. Third-trimester uterine rupture without previous cesarean: A case series and review of the literature. Am J Perinatol 2009;26:739–44. doi:10.1055/s-0029-1223287.

[2] Ofir K, Sheiner E, Levy A, Katz M, Mazor M. Uterine rupture: differences between a scarred and an unscarred uterus. Am J Obstet Gynecol 2004;191:425–9. doi:10.1016/j.ajog.2004.01.026.

[3] Walsh CA, Baxi LV. Rupture of the primigravid uterus: a review of the literature. Obstet Gynecol Surv 2007;62:327–34 quiz 353-354. doi:10.1097/01.ogx.0000261643.11301.56.

[4] Gibbins KJ, Weber T, Holmgren CM, Porter TF, Varner MW, Manuck TA. Maternal and fetal morbidity associated with uterine rupture of the unscarred uterus. Am J Obstet Gynecol 2015;213 382.e1-6. doi:10.1016/j.ajog.2015.05.048.

[5] Divon MY, Haglund B, Nisell H, Otterblad PO, Westgren M. Fetal and neonatal mortality in the postterm pregnancy: the impact of gestational age and fetal growth restriction. Am J Obstet Gynecol 1998;178:726–31. doi:10.1016/s0002-9378(98)70482-x.

[6] Ingemarsson I, Källén K. Stillbirths and rate of neonatal deaths in 76,761 postterm pregnancies in Sweden, 1982–1991: a register study. Acta Obstet Gynecol Scand 1997;76:658–62. doi:10.3109/00016349709024606.

[7] Caughey AB, Musci TJ. Complications of term pregnancies beyond 37 weeks of gestation. Obstet Gynecol 2004;103:57–62. doi:10.1097/01.AOG.0000109216.24211.D4.

[8] Revicky V, Muralidhar A, Mukhopadhyay S, Mahmood T. A case series of uterine rupture: lessons to be learned for future clinical practice. J Obstet Gynaecol India 2012;62:665–73. doi:10.13224/012-0328-4.

[9] Abebe F, Mannekulih E, Megerso A, Legese T. Determinants of uterine rupture among cases of Adama city public and private hospitals, Oromia, Ethiopia: a case control study. Reprod Health 2018;15:161. doi:10.1186/s12978-018-0606-4.

[10] Lydon-Rochelle M, Holt VL, Easterling TR, Martin DP. Risk of uterine rupture during labor among women with a prior cesarean delivery. N Engl J Med 2001;345:3–8. doi:10.1056/NEJM200107053450101.

[11] Kotoulová M, Mikysková I, Drášková J, Vláčil J, Dvořák M, Haška M. Adrenocortical oncocytoma presenting as Cushing’s syndrome in pregnancy with spontaneous postpartum uterine rupture. Ceska Gynelk 2016;81:228–32.

[12] Favilli A, Pericoli S, Acanfora MM, Bini V, Di Renzo GC, Gerli S. Pregnancy outcome in women aged 40 years or more. J Matern Fetal Neonat Med 2012;25:1260–3. doi:10.3109/14767058.2011.643327.

[13] Ziadah S, Yahaya A. Pregnancy outcome at age 40 and older. Arch Gynecol Obstet 2001;265:30–3. doi:10.1007/s0044400000122.

[14] Hsieh TT, Liou JD, Hau JJ, Lo LM, Chen SF, Hung TH. Advanced maternal age and adverse perinatal outcomes in an Asian population. Eur J Obstet Gynecol Reprod Biol 2010;148:21–6. doi:10.1016/j.ejogrb.2009.08.022.