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Case Report

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

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ABSTRACT

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. Suppuration 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.

Abbreviations: AMA, Advanced maternal age; CT, Computed tomography; MRI, Magnetic Resonance Imaging.

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

Introduction

Uterine rupture is a rare and life-threatening complication for both mother and child [1,2]. It usually occurs in scarred uteri 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.

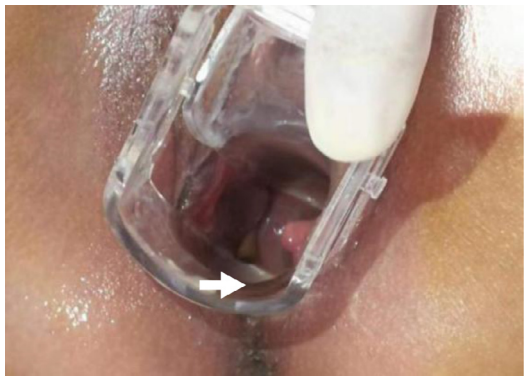

Fig 2 – Gynecological examination showed a fetal bone at the orifice of the cervix (white arrow).

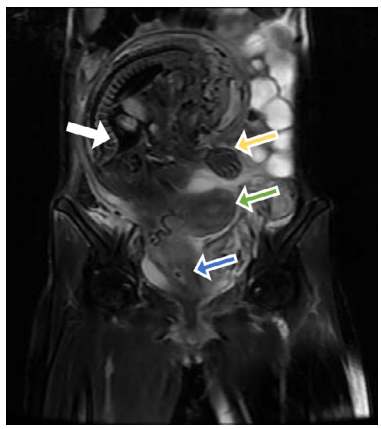

Fig 3 – Coronal MRI showed the fetus was in breech position, and uterus was enlarged, especially at the right uterine horn (white arrow). Uterine wall is shown with green arrow. Fetal hand protruded into patient abdominal cavity (yellow arrow), and fetal femur bone protruded into the vagina (blue arrow). (Color version of figure is available online)

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

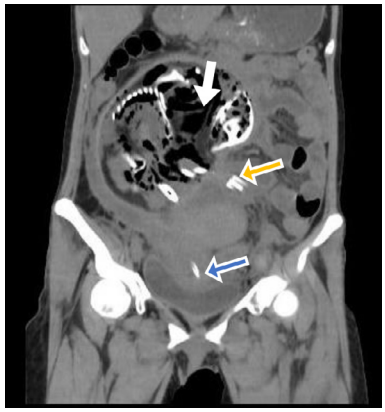

Fig 4 – Coronal CT revealed considerable amount of gas was found in the uterus (white arrow), fetal hand protruded into patient abdominal cavity, and fetal femur bone (yellow arrow) protruded into the vagina (blue arrow). (Color version of figure is available online)

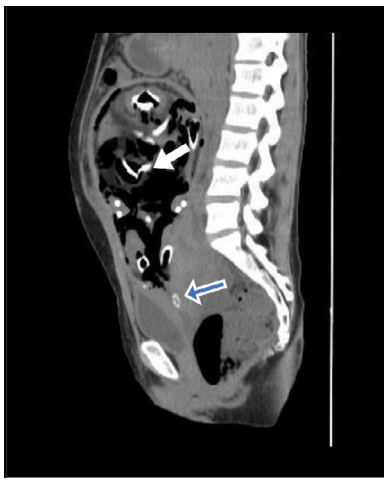

Fig. 5 – Sagittal CT revealed considerable amount of gas in the uterus (white arrow). Fetal femur bone of the deceased fetus protruded into the mother's vagina (blue arrow). (Color version of figure is available online)

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.

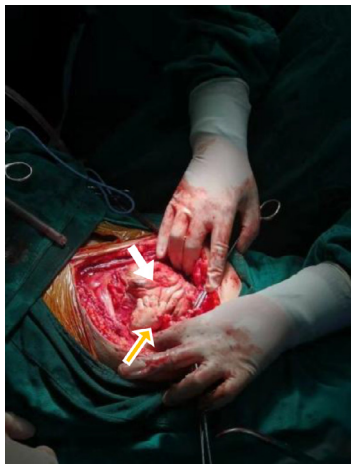

Fig 6 – Thick amniotic sac formed a cystic wall surrounding the fetus, adhered closely to the abdominal wall, omentum, some small bowels, and retroperitoneum (white arrow), with pus and odor present The right fetal hand protruded into the amniotic sac (yellow arrow). (Color version of figure is available online)

Fig 7 – Fetus was 20 cm long and fetal head was seriously infected (green arrow). The tissue below the two knee joints of the fetus was lost (white arrow). (Color version of figure is available online)

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. 7,8). The patient's ovaries and fallopian tubes were severely swollen and deformed. As a result, hys-

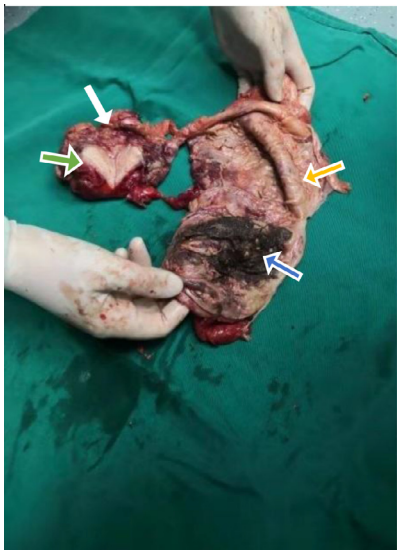

Fig 8 – The uterus (green arrow) and ruptured uterine scar (white arrow). Significantly thickened amniotic sac (yellow arrow). Necrotic fetal hair (blue arrow). (Color version of figure is available online)

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

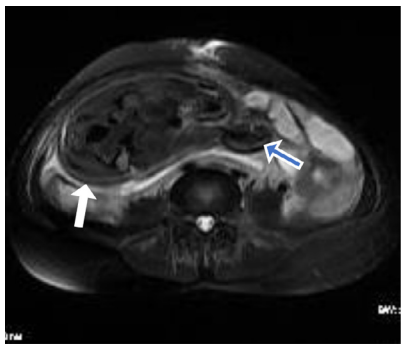

Fig 9 – Axial MRI: radiologists mistakenly regarded the amniotic sac wall as the uterine wall of right cornual pregnancy (white arrow). Fetal hand protruded into the patient's abdominal cavity (blue arrow). (Color version of figure is available online)

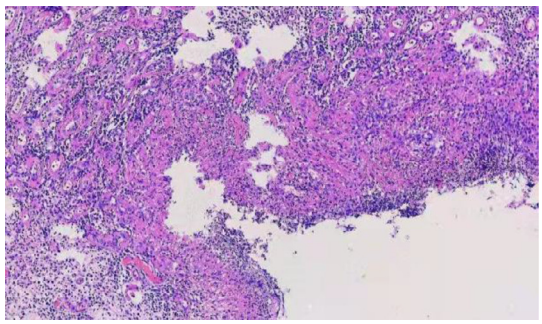

Fig 10 – . Pathology: focal abscess formed in the myometrium and serosa, and granulation tissue proliferated. Diffuse infiltration of lymphocytes and plasma cells in the stroma of bilateral tubal mucosa.

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

congenital breech position of the fetus provided a channel at the cervix via the vagina. Some suppuration and bone tissue discharge relieved the pressure in the abdominal cavity and prevented the occurrence of severe peritonitis.

Aging is associated with an increased prevalence of maternal medical disorders that may adversely impact the health of pregnancy [12]. Advanced maternal age (AMA) women have increased rates of hypertensive disorders, diabetes, placental abruption, and abnormal placental locations [13,14]. 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.

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