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CASE REPORTS AND LITERATURE REVIEWS

Uterine rupture in nulliparous woman without risk factors: a case report and literature review

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Objective: The aim of this study was to summarize the main features of spontaneous uterine rupture in primigravid patients before the onset of labor, emphasize the possibilities of therapeutic conduct, and offer points of reflection on the post-rupture management.
Methods: We performed a literature review of all the individual case reports, retrospective case series, and reviews concerning uterine rupture in peer-reviewed journals from January 1975 to October 2021.
Results: The diagnosis of uterine rupture was commonly made by abdominal pain, with or without concomitant nausea and vomiting. Uterine ruptures occur more frequently at the end of pregnancy or in the third trimester. The most frequently involved site is the uterine cornua followed by the posterior wall of the uterus. Other described cases identify the broad ligament, uterus sacral ligament, lower uterine segment, and anterior wall as possible points of rupture. The most frequently used suturing technique is the repair of the breach in two layers.
Conclusion: Uterine rupture is an extremely rare obstetric emergency, correlated to life-threatening consequences for both the newborn and the woman. Considering the maternal and fetal risks, a tempestive diagnosis is mandatory.

Keywords: uterine rupture, UR, unscarred uterus, nulliparous, primigravid

Introduction

Uterine rupture (UR) is an extremely rare and unexpected obstetric complication. It consists of a full-thickness tearing of the three uterus layers (endometrium, myometrium, and perimetrium).

Normally, it occurs in women who have undergone surgery, typically a caesarian section; nevertheless, a small number of cases occur in unscarred uteri (1).

In developed countries, the incidence of rupture, in the absence of previous cesarean delivery, is estimated to be 1/15,000–1/20,000 pregnancies (1–6).

In less developed countries, UR remains a frequent and important cause of maternal mortality and morbidity because prompt access to cesarean section and operative vaginal delivery for the management of intrapartum complications is not readily available (7, 8). In a recent review of 34 cases, Khan et al. reported an incidence of UR in a single center in Pakistan of 1/100 deliveries (9). Furthermore, it has been found that multi-parity, in particular grand multi-parity, and contracted pelvis, whose incidence is higher in black women, constitute an important risk factor of rupture (8, 10, 11).

Another element that must be carefully evaluated when collecting the patient's medical history is the presence of a surgical scar at the level of the lower uterine segment, which is usually caused by a previous cesarean section. Other factors taken into consideration are surgical manipulations of any kind (i.e., curettage, manual removal of the afterbirth), trauma, mal-presentation, great multi-parity, use of uterotonic agents, abnormal placentation, and Mullerian anomalies (4, 7–17).

Clinical presentations of UR expose acute abdominal pain, fetal heartbeat abnormalities (bradycardia, occasionally associated with late deceleration), vaginal bleeding, abdominal pain during labor, uterine tenderness and change in uterine shape, and hematuria in case of bladder lesions (4–6, 13, 18–20).

The role of diagnostic imaging in this case is not decisive (21). If the patient is stable, the ultrasound examination is recommended, and it may identify free fluid in the abdomen or uterine wall's alterations, which can raise the suspicion of UR (13, 22). In most cases, the diagnosis is made by laparotomy; this procedure allows to identify the area affected by the rupture and the cause of the bleeding. By virtue of the extreme rarity of this event, only a few cases involving women in their first pregnancy are reported in the literature. We were able to find only 15 cases of UR in the absence of established risk factors.

The aim of this review was to summarize the main features of spontaneous UR in primigravid patients before the onset of labor, emphasize the possibilities of therapeutic conduct, and offer points of reflection on the post-rupture management.

Concurrently, we report a case of spontaneous rupture in a nulliparous patient at her 21st week of gestation with no apparent risk factors occurring in our center.

Methods

We have performed research in the literature in PubMed, Embase, Cochrane Library, and MEDLINE, starting with individual case reports, retrospective case series, and reviews concerning UR in peer-reviewed journals. The language was restricted to English. The publication range was from January 1975 to October 2021. We used the keywords "uterine rupture" in "primigravid/nulliparous/first pregnancy/without risk factors" in combination with "unscarred/without risk factor" and "before on set labor/outside labor/pre labor." This review examines documented cases of spontaneous UR in primigravid women. Ruptures secondary to trauma and incomplete ruptures are not included.

Results

Case report

A 31-year-old Caucasian woman in her first pregnancy was admitted at 21 weeks of gestation with lower abdominal pain associated with nausea and vomiting. Her antenatal course had been uneventful. There was no history of vaginal bleeding, rupture of membranes, uterine contractions, abdominal trauma, previous pelvic surgery, or drug abuse. She said she had suffered from severe abdominal pain and later lost consciousness. Clinically, on examination, she was distressed, pale, tachypnoeic, and hypotensive (blood pressure was 100/60). The abdominal examination revealed an increment of uterine tachtone, which was tetanic. No rebound tenderness was found. The vaginal examination showed a closed cervix with no discharge. The ultrasound reported a singleton fetus with normal biometry, the placenta was posterior-fundal with no signs of abruption, and the amniotic liquid was regular. A severe fetal bradycardia of 65 bpm was found on the ultrasound evaluation. Subsequently, an immediate resuscitation with colloids was started. Blood chemistry tests were normal, with a hemoglobin level of 12 g/dL. In consideration of the tetanus consistency of the uterus and fetal bradycardia, we started the intravenously tocolysis.

After 2 h, on the re-evaluation, the woman appeared pale but with a normal blood pressure (BP) value, the fetal heart rate (FHR) had returned to a normal range, and the uterus appeared released.

Six hours after the admission, she deteriorated with hypotension (BP of 90/60), tachycardia, and tachypnea. She complained about increased abdominal pain and difficulty breathing. Her blood tests showed a decrease in hemoglobin levels (Hb 10 g/dL). An abdominal-pelvic computed tomography (CT) was performed, and a diagnosis of hemoperitoneum was posed. An emergency laparotomy was performed under general anesthesia. A massive hemoperitoneum was detected. A high abdomen as a cause of bleeding was excluded. An evaluation of the uterus and adnexa showed profuse bleeding caused by a laceration of the posterior wall of the uterus, which extended to the parametrium, left round, and uterosacral ligaments. An attempt of conservative management was essayed. Considering the impossibility to control the hemorrhage, the surgeons decided to perform a cesarean section with the aim of terminating the pregnancy, reducing the uterine volume and its blood perfusion. At the end of the pregnancy termination, the bleeding was reduced thanks to hemostatic suture. Total blood loss was approximately 2,700 mL. Postoperative course required blood and plasma transfusions. The patient progressively recovered and was discharged 10 days after surgery. The subsequent follow- up was regular.

Results

In our review, we identified 15 cases of UR in primigravid women, as reported in **Table 1**, including the one presented in this study by our group, apparently without identified risk factors.

The average age of the women in this article is 26.4 years. The diagnosis of UR was most commonly made by abdominal pain, with or without concomitant nausea and

TABLE 1 | Antepartum uterine rupture in the unscarred uterus in a primigravid woman without apparent cause.

Author, reference	Year	Age	GA	Presenting symptoms	Rupture site	Fetal outcome	Treatment
Nel (31)	1989	19	38	Sudden onset of acute abdominal pain. Fetal tachycardia of 180/min	Posterior uterine wall	Live birth	Repaired in two layers
Fischer (32)	1996	16	30	Lower abdominal pain. A fetal heart rate tracing was reactive, and no vaginal bleeding	Posterior wall extending toward the cervix	N/A	Was repaired in layers
Langton (21)	1997	27	32	Sudden sharp abdominal pain, nausea shoulder pain and slight difficulty in breathing. The fetus heart rate was normal	The right uterosacral area	Live birth	Repair without tubal
Matsubara (33)	2011	27	38 + 6	Weak abdominal pain	Uterine anterior wall	Live birth	Excised the thin part of the uterine wall and reconstructed the site
Mishina (34)	2014	36	32	Severe abdominal pain and reduced FM, no vaginal bleeding	A horizontal incision was made in the uterine lower segment	Live birth	Repaired
Wang (35)	1999	30	40	Abdominal pain and fetal distress	Cornual site	Live birth	Repaired double layer
Wang (29)	2006	26	30	Fetal distress (severe variable deceleration)	No mention	Live birth	Repair without tubal sterilization
Wang (29)	2006	30	40	Fetal distress (severe variable deceleration) Abdominal pain with peritonitis sign	No mention	Cerebralpalsy	Repair without tubal sterilization
Abbi and Misra (36)	2002	20	37	Abdominal pain; loss of FM	Left cornual area	Stillbirth	Repaired
Mizutamari (16)	2014	29	31	No symptoms	Right cornual area	Live birth	Interrupted vicryl sutures
Zhao (37)	2017	25	36 + 2	Abdominal pain	Broad ligament	Live birth	Repaired in one layer
Hawkins (30)	2018	35	21 + 1	Abdominal pain	Fundal	Live birth	Repair without tubal sterilization at 21 weeks and elective caesarean section due to premature rupture of amniotic sac at 32 sg
Yang (38)	2021	35	27	Mild abdominal discomfort and oligoamnios	Right uterine cornua	Live birth	With several figure-of-eight sutures
Katwal (39)	2021	25	11	Abdominal pain and vomiting	Cornual site	Stillbirth	Repaired in double layers
Our case	2021	33	21	Abdominal pain and vomiting	Posterior uterine wall	Stillbirth	Repaired in double layers

GA: gestational age at rupture (weeks); Nil: no mention; Lt: left; Rt: right; FM: fetal movement.

vomiting. Of those women who underwent labor, they were diagnosed with abnormal FHR.

There were no maternal deaths.

As already reported in the literature, UR, although spontaneous, occurs more frequently at the end of pregnancy or in the third trimester. In the series we assessed, 2/15 women were in the second trimester of pregnancy, 5/15 at the end of pregnancy, and the remaining in the third trimester (between the 27th and 36 + 2 weeks) (1–3).

We observed that the site that is most frequently involved in the rupture is the uterine cornua (5/15 cases), followed by the posterior wall of the uterus (3/15). Other described

cases identify the broad ligament, uterus sacral ligament, lower uterine segment, and anterior wall as possible points of rupture (3, 23, 24).

Fetal outcomes, where reported, show 12/15 newborns alive and viable at birth, 1/15 complicated by cerebral palsy, while 2/15 fetal deaths were described.

The most frequently used suturing technique is the repair of the breach in two layers. No mention is made of the type of thread or needle used.

Few data were reported in the literature about pregnancy outcomes after UR out of labor (13, 23–25); to the best of our knowledge, we only found a subsequent letter to the editor by Walsh, which revealed that the same woman who suffered from UR in his case report carried another pregnancies with a spontaneous birth at term (26). Moreover, the possibility of UR recurrence after previous rupture of a low-lying C-section, during labor, has an estimated incidence of 5% (27). No information was described in patients with other sites of uterine rupture, but we considered a similar or higher risk.

Discussion

UR is a rare obstetric emergency to keep in mind, due to its extreme and life-threatening consequences for both the newborn and the woman (2–4).

Due to the infrequency of the phenomenon and the low quality of the evidence circulating in the literature, making a timely diagnosis can be extremely difficult. Considering the maternal and fetal risks, a tempestive diagnosis is mandatory; therefore, we decided to report our case and make a literature review that will help those who will face this situation in the future.

In the literature, it is easy to find a large number of cases of UR, occurring during labor, in women who had undergone previous surgery, especially a cesarean section (1, 12, 13, 15, 16).

On the contrary, spontaneous ruptures, in women at their first pregnancy, are undoubtedly rarer (13, 22).

Therefore, the peculiarity of our case coincides with an early gestational age at the onset; specifically, this is one of only two cases reported in the literature in the second trimester of pregnancy, without a risk factor. In contrast, this complication usually occurs in more advanced pregnancy, in particular in 5/14 at the end of the pregnancy and 7/14 in the third trimester, as reported in our literature review (23, 24).

In general, the symptomatology is not extremely characteristic and could limitedly support the clinician in the diagnosis. From our review, the only symptom that must be recognized as a potential red flag at any gestational week is acute abdominal pain associated with hemorrhagic shock or vasomotor symptoms (17–20).

Imaging, as our case confirmed, plays a limited role in identifying this condition; the ultrasound scan is not nullifying in the management of UR. Even the CT scan only points out the hemoperitoneum without showing a solution of continuity; in addition, it can only be performed in a stable patient, which is not so common in case of massive hemoperitoneum. For these reasons, unfortunately, the diagnosis has to be made through a surgical approach. Thus, the indication is to stabilize the patient, starting with an ultrasound examination and then evaluate the necessity of other imaging; however, in case of suspicion or absence of other causes that can explain the clinical condition, a surgical examination is required (20, 28).

Although the diagnostic process is extremely challenging, there was no maternal death or C-section with peripartum hysterectomy in this review. So, we can assume that, in these cases, it is easier to manage conservatively.

The only two fetal deaths reported in this series refer to our case, at a gestational period when the fetus is still incompatible with life, and Wang, of which no further details are reported (29). Hawkins et al. reported a case of a fundal complete uterine defect (10 cm) with protrusion of the chorioamniotic membrane, in the midtrimester at 21 weeks, which at the behest of the patient was not interrupted, but a double layer repair of the area was carried out, and then the woman gave birth by cesarean section at 32 + 1 gestational weeks (30). This is probably the only similar case to the one presented by us, which involved management diametrically opposite to that carried out by our center. Unfortunately, the frequency of UR is too low to decide univocally what must be done. In particular, although a conservative management could be evaluated, in our case, the impossibility to control the hemorrhage led the surgeon to prefer a termination of pregnancy. Therefore, the individual situations should be evaluated case by case, according to the maternal symptoms and the uterine repair capabilities, as well as the monitoring of the bleeding.

The issue of UR in women with their first pregnancy should be taken into consideration as a point of reflection for future pregnancies; specifically, accurate and multidisciplinary counseling must be performed considering the high risk of recurrence associated with maternal and fetal morbidity and mortality. To the best of our knowledge, only Walsh et al. reported a subsequent pregnancy without complications, after a previous UR. No other cases were reported (26). In particular, not even Walsh reported on the management of these women once they have undergone a second pregnancy. Even if this case is reassuring, considering the possibility of subsequent pregnancy, there is a limit imposed by the lack of information to manage an adequate follow-up of a patient interested in a procreative future.

Conclusion

UR is an extremely rare obstetric emergency with life-threatening consequences for both the new born and the woman (2–4).

Due to the infrequency of the phenomenon and the low quality of the evidence circulating in the literature, making a timely diagnosis can be extremely difficult. Considering the maternal and fetal risks, a tempestive diagnosis is mandatory; therefore, we decided to report our case and make a literature review that will help those who will face this situation in the future.

We would like to underline the following:

There are no typical signs and symptoms, even though an UR must be taken into consideration in the case of acute abdominal pain associated with hemorrhagic shock.

1. Imaging is not diagnostic, and in suspected cases a surgical approach is mandatory.
2. Future studies must be implemented to evaluate the possibility of conservative management; at the moment, the best approach is to evaluate case by case considering the individual situation, monitoring maternal symptoms, the uterine repair capabilities, and the bleeding's extent.
3. Another challenging issue is the counseling for couples that desire a future pregnancy after a previous UR: no data were reported in the literature about the correct management and frequency of pregnancy evaluations, while there is an elevated risk of recurrence.

Author contributions

All authors contributed equally to the manuscript and read and approved the final version of the manuscript.

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