Case Report
Spontaneous prelabor unscarred uterine rupture in primigravida: a case report and review of literature

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Abstract: Spontaneous uterine rupture (SUR), a public health problem in developing countries, is a rare but hazardous obstetric complication with significant maternal and fetal morbidity and mortality. Cases of rupture are more likely to occur in a scarred uterus, usually after a previous caesarean delivery. Spontaneous unscarred uterus rupture (SUUR) in primigravid women is usually unexpected and extremely rare. Risk factors for SUUR include high parity, placental abnormalities, and uterine anomaly. Hereby, we presented a case of spontaneous unscarred uterus rupture during her pregnancy of 36 weeks +2 days in a primigravid woman without apparent causes. We then performed literature review covering similar cases to further remind physicians of the possibility of SUUR.

Keywords: Spontaneous, prelabor, unscarred uterus, rupture, primigravid

Introduction

Uterine rupture is a rare but hazardous obstetric complication with catastrophic outcomes. Although much progress has been made, uterine rupture is still a life-threatening obstetric emergency bearing a high risk for the mother and the fetus, especially in developing countries [1-3]. Compared to the mortality of mothers, it is more hazardous to fetuses [4]. If the uterine rupture develops in an unscarred uterus, the prognosis is even worse [2]. On the other hand, it is rarely seen. The overall incidence of uterine rupture is as low as 1 in 1,146 pregnancies (0.07%) even in high-risk subgroups, according to pooled data from meta-analysis of 25 studies in 1976-2012 [5].

Several risk factors are associated with uterus rupture, but the most common one is the previous cesarean section [6]. Rupture in an unscarred uterus is especially rare. The primigravid uterus is considered immune to spontaneous rupture [7]. However, rare cases still exist. Cases alike in this study are somewhat insidious at the onset of disease, which lack specific symptoms and finally lead to a misdiagnosis or a delayed diagnosis. Therefore, prompt and correct diagnosis followed by definitive treatment plays an important role in reducing morbidity and mortality.

In this article, with no apparent causes, a case of spontaneous unscarred uterus rupture in a primigravid woman before labor was reported and a literature review was performed.

Case report

A 25-year-old primigravid woman at 36+2 weeks of gestation was sent to our obstetric emergency department with a complaint of acute abdominal pain for two hours. She had no antepartum hemorrhage or vomiting. Her past history was unremarkable. Routine pregnancy check had been performed at our hospital for 9 times from the early gestation ultrasound scan at the 20th week of gestation, all demonstrating that the pregnancy was normal.

The day before her admission, she visited our department because of mild lower abdominal pain. When she arrived at the hospital, the abdominal pain was significantly alleviated. Ultrasonography revealed the pregnancy was...
SUUR in primigravida: case report and literature review

intrauterine normal. The cardiotocograph was performed and the episode was settled spontaneously.

After returning home, she felt well until approximately 2 hours before the next admission when she sensed a sudden onset of acute abdominal pain (self-rated fetal heart rate >120 bpm regular). On her way to the hospital, she felt that the pain was getting worse and spread throughout her entire abdomen. Yet she did not sense any contractions of labor.

On admission, she was pale and distressed with a PR 104/min and BP 100/70 mmHg. Abdomen was distended and tense, but no rebound tenderness was found. Because of the tense uterus, the fetus was difficult to feel. Speculum examination revealed an upper positioned cervix and no vaginal bleeding. On digital examination, the cervix was tightly closed, the fetus was untouched. A cardiotocograph showed a prolonged fetal bradycardia with 65 bpm no reaction. Although the clinical picture was atypical, there was still possibility of abruption placenta because of the absence of blood in the amniotic fluid (the fetal presenting part was high and not well applied to the cervix, so artificial rupture of fetal membranes is difficult) and thus a definitive uterine rupture could also be possible. Due to the seriously deteriorating state of the fetus, an emergency laparotomy was performed under lumbar anesthesia.

After entering the peritoneal cavity, approximately 2 L blood and fresh clots were drained. The whole intact amniotic sac and the fetus were found floating in the abdominal cavity. A 2700 g male fetus were extracted with no need for hysterotomy. The amniotic fluid was clear and a large complete laceration was found to extend vertically, from the left of the midline throughout the whole broad ligament. The inferior extremity reached the cervical OS with the upper extremity over the left fundus, ending in the right corner. The placenta was still attached in anterior position without any evidence of abruption or accrete (Figure 1). The uterus showed no structural abnormalities and both fallopian tubes and ovaries were normal.

Considering the patient had no child delivery before, we repaired the uterine with a continuous double-layer closure using 1-0 synthetic interrupted vicryl I without undertaking tubal ligation. The patient was transfused with a 4 U red cell pack and a 2 U fresh frozen plasma. The infant’s Apgar scores were 1 at 1 min, 5 at 2 min, 4 at 10 min, cerebral palsy was diagnosed. The patient was discharged on her 8 postoperative day and was advised to avoid further pregnancy for at least two years.

Discussion

Incidence, etiology, pathogenesis and literature sources

Incident rate of pregnancy-related uterine rupture is 1/1146 pregnancies and estimated incidence of spontaneous rupture of unscarred uterine (SUUR) was 1/8,434 pregnancies (0.012%) in industrialized countries and 1/920 pregnancies (0.11%) in developing countries [5]. A 10-year Irish study by Gardeil et al. [8] showed that the overall incidence rate of unscarred uterine rupture during pregnancy was 1 in 30,764 deliveries (0.0033%). No uterine rupture occurred among 21,998 primigravidas and only 2 (0.0051%) occurred among 39,529 multigravidas with no uterine scar. As Kaczmarek et al. [9] stated in 1994 that “the overall risk of uterine rupture in the primigravid patient was thought to be negligible”, the obstetric event is extremely rare.

Despite the relative rarity of this phenomenon, this obstetric emergency is life threatening to mother and fetus. Therefore, investigating its etiology and pathogenesis is of significant importance.

Rupture of the pregnant uterus not only occurs due to the existed damage or defect, but could also in an unscarred uterine. The unscarred uterine rupture (UUR) could be either traumatic or spontaneous. Traumatic factors include pen-
SUUR in primigravida: case report and literature review

...tering abdominal trauma, labor induction, and in particular the usage of oxytocin or prostaglandin [4, 5]. An internal podalic version, breech extraction, and instrumental delivery are also associated with traumatic rupture [10]. Multiparity, particularly grand multiparity, leading to weakened muscle fibers and scarring from previous labors and deliveries as well as uterine muscular weakness, is the most common risk factor for spontaneous unscarred uterine rupture (SUUR) [5, 11]. Other risk factors include placental abnormalities and adenomyosis [7].

Identified unscarred uterus rupture (UUR) in primigravida risk factors include obstructed labor, congenitally abnormal uterus, fetal malpresentation, and Ehlers-Danlos syndrome (EDS) [7, 12].

Unscarred uterus rupture in primigravida with no apparent cause is an exceedingly rare event. Walsh and Baxi [7] reviewed the literature over six decades (1946-2006) and found that among 36 primigravid uterine ruptures, only 2 had unknown risk factors, and most cases had at least one risk factor. Waters and Hall stated that, “rupture of the uterus of a primigravida in late pregnancy without a discernable cause is one of the rarest obstetric oddities” [13].

Despite of the widely held belief that the primigravid uterus reacts to obstruction with uterine inertia, few studies reported that the only identifiable factor for primigravid uterine rupture is labor-related uterine contraction [14].

Although uterine ruptures of all types were reviewed, our primary interest focused on rare, unusual and spontaneous uterine ruptures from 28 weeks of pregnancy to the onset of labor without an apparent cause. There are only 8 similar cases documented in literature. Among them, 5 were found in individual case reports [15-19], 2 were found in retrospective case series [10], 1 from further references researching [20]. This case was referred by Etsuko [21] and Uccella [22]. In a case series, 39 cases of uterine rupture were reported from 114,933 deliveries [23]. Three occurred in the unscarred uterus and one was primigravida with no history of uterine surgery. Other threatening factors such as whether rupture occurred in antepartum was not mentioned. In addition, Walsh CA, et al. [24] reported an unexplained primigravid uterine rupture, while following skin biopsy confirmed the diagnosis of EDS type IV in a subsequent pregnancy [12]. This case was not included in our review (Table 1).

Classification

Uterine rupture is the uterine wall tearing during pregnancy or delivery. Based on anatomy and etiology, it can be categorized into two types, namely, complete rupture and incomplete rupture [25]. These two types are distinct in that the former does not extend the entire thickness of the uterus which is covered by peritoneum. It often gradually occurs in the area scarred from the previous uterine, with little hemorrhage, the fetus is not extruded, and generally is painless. Comparatively, complete rupture is a much worse event resulting in direct communication between the uterine and peritoneal cavities [26], with extrusion of intrauterine contents into the peritoneal cavity. It often occurs in an unscarred uterus [3]. Compare with incomplete rupture, it usually occurs suddenly, and is often associated with severe pain and signs of significant blood loss.

In the case of SUUR in primigravida without identified risk factors, 50% (n=4) was found to have complete rupture while 25% (n=1) was incomplete and 37.5% (n=3) was “no mention”. Without considering the final uterine feature, it remains to be verified whether all or several complete ruptures in primigravid uterus progresses through incomplete ruptures. In some cases, complete rupture is the result, while incomplete rupture is the process. This is identical with what has been verified in our case. The amniotic fluid was found clear in laparotomy, while the adherent section of placenta was not at the metrorrhaxis places. The time when the metrorrhaxis occured could not be confirmed since there was a high possibility of rupture at home or on the way to hospital.

Clinical presentation and diagnosis

The typical signs and symptoms of uterine rupture in the third trimester are the sudden occurrence of severe and shearing abdominal pain with cessation of uterine contractions while vaginal bleeding and shock occurs. The fetus suffers in utero distress with bradycardia as well as decreased fetal movement. The infrequent symptoms of uterine rupture are epigas-
## Table 1. Antepartum uterine rupture in the unscarred uterus in a primigravid women without apparent cause

<table>
<thead>
<tr>
<th>Author, Reference</th>
<th>Age</th>
<th>GA</th>
<th>Presenting symptoms</th>
<th>Rupture site</th>
<th>Type of rupture</th>
<th>Fetal weight (g)</th>
<th>Apgar score 1 min</th>
<th>Apgar score 5 min</th>
<th>Fetal outcome</th>
<th>Treatment</th>
</tr>
</thead>
<tbody>
<tr>
<td>Nel JT [15]</td>
<td>19</td>
<td>38</td>
<td>Sudden onset of acute abdominal pain. Fetal tachycardia of 180/min</td>
<td>Posterior uterine wall</td>
<td>Complete</td>
<td>2822</td>
<td>5</td>
<td>9</td>
<td>Live birth</td>
<td>Repaired in two layers</td>
</tr>
<tr>
<td>Fischer RL [16]</td>
<td>16</td>
<td>30</td>
<td>Lower abdominal pain. A fetal heart rate tracing was reactive, and no vaginal bleeding</td>
<td>Posterior wall extending toward the cervix</td>
<td>Complete</td>
<td>1080</td>
<td>3</td>
<td>8</td>
<td>Live birth</td>
<td>Was repaired in layers</td>
</tr>
<tr>
<td>Langton J [17]</td>
<td>27</td>
<td>32</td>
<td>Sudden sharp abdominal pain, Nausea shoulder pain and slight difficulty in breathing. The fetus heart rate was normal</td>
<td>The right utero sacral area</td>
<td>Complete</td>
<td>2040</td>
<td>2</td>
<td>7</td>
<td>Live birth</td>
<td>Repair without tubal</td>
</tr>
<tr>
<td>Matsubara S [18]</td>
<td>27</td>
<td>38+6</td>
<td>Weak abdominal pain</td>
<td>Uterine anterior wall</td>
<td>Incomplete</td>
<td>2604</td>
<td>6</td>
<td>8</td>
<td>Live birth</td>
<td>Excised the thin part of the uterine wall and reconstructed the site</td>
</tr>
<tr>
<td>Mishina M [19]</td>
<td>36</td>
<td>32</td>
<td>Severe abdominal pain and reduced FM, no vaginal bleeding</td>
<td>A horizontal incision was made in the uterine lower segment</td>
<td>Complete</td>
<td>1807</td>
<td>6</td>
<td>8</td>
<td>Live birth</td>
<td>Repaired</td>
</tr>
<tr>
<td>Wang YL [10]</td>
<td>30</td>
<td>40</td>
<td>Fetal distress (severe variable and late deceleration)</td>
<td>No mention</td>
<td>Nil</td>
<td>3182</td>
<td>4</td>
<td>9</td>
<td>Live birth</td>
<td>Repair without tubal sterilization</td>
</tr>
<tr>
<td>Wang YL [10]</td>
<td>26</td>
<td>30</td>
<td>Fetal distress (severe variable deceleration) Abdominal pain with peritonitis sign</td>
<td>No mention</td>
<td>Nil</td>
<td>1610</td>
<td>1</td>
<td>2</td>
<td>Cerebral palsy</td>
<td>Repair without tubal sterilization</td>
</tr>
<tr>
<td>Abbi and Misra [20]</td>
<td>20</td>
<td>37</td>
<td>Abdominal pain; Loss of FM</td>
<td>Left cornual area</td>
<td>Nil</td>
<td>Nil</td>
<td>Nil</td>
<td>Nil</td>
<td>Live birth</td>
<td>Repaired</td>
</tr>
</tbody>
</table>

GA: gestational age at rupture (weeks), Nil; no mention, Lt; left, Rt; right, FM; fetal movement.
tric pain [27], shoulder pain (right sided or bilateral) [28], abdominal distention and paralytic ileus as well as, hematuria, etc. However, it is rare to find these symptoms prenatally especially in the rupture of the unscarred uterus.

Literature review showed eight cases of SUUR of primigravida. In these eight cases, three presented severe abdominal pain (37.5%), one presented weak abdominal pain (12.5%) and the other three reported no abdominal pain degree. No intrauterine fetal demise was found. Three (37.5%) was found with fetal compromise while two with normal fetal heart rate, the other three cases lack description for fetal heart rate. Fetal movement was reduced in one case while lost in another one. Other six cases lack the description for fetal movement. Vaginal bleeding was not a reliable symptom due to the fact that none of these patients was found with vaginal bleeding or shock in our review. Moreover, since all the uterine ruptures occurred prior to the onset of labor, there was no uterine constriction. One (25%) was found with nausea, shoulder pain and slight difficulty in breathing. None of the patients was found with hematuria or other specific signs.

Although the presenting features are similar to those in a parous patient, most physicians have a lower index of suspicion for primigravid rupture. This means that interval to surgical intervention could be delayed in primigravidas, especially if the diagnosis is not initially considered. Unscarred uterus would be worse as the area of rupture is rich in vessels [1, 5]. Although there was no confined weak area, such as a previous incision, the tear may still progress and involve vital, adjacent organs.

Diagnosis and antidiastole

A number of conditions can present with abdominal pain, hypovolemia, and fetal compromise in the primigravid women. Most commonly, this constellation occurs with placental abruption, which could be concealed in the absence of vaginal bleeding. As in our case, initial diagnosis was placental abruption. Other less common conditions to be considered include subcapsular liver hematoma with or without rupture, rupture of the broad ligament [29], splenic rupture, appendicitis, biliary colic, pancreatitis, peptic ulcer disease, intestinal obstruction. Urinary tract disorders, ovarian tumors or torsion, uterine torsion [30] and uterine vein rupture [31] are also included.

After a careful history and physical examination, laboratory tests including type and cross match for blood, complete blood count, electrolytes, blood urea nitrogen, creatinine, and urinalysis should be carried out. In patients with right upper quadrant pain, liver enzymes and serum amylase also may be helpful. Necessary auxiliary examination and therapy shall not be delayed because the fetus is still in the abdominal cavity. Ultrasonography is probably the safest and most useful imaging technique during pregnancy. Intrauterine blood, dissociating peritoneal blood, empty uterus, and large uterine mass with gas bubbles have been reported as sonographic findings associated with uterine rupture.

Obstetric consultation in all pregnant women with abdominal pain must be considered. In addition, if the pregnancy is reaching or near full-term, fetal health condition can be inspected through ultrasonography and fetal monitoring.

Once the uterine rupture is considered, instant laparotomy is generally indicated for a patient presenting with these symptoms.

Site of rupture

According to the findings of Donnelly [32] and Birger [33], the left lateral wall of the uterus is more prone to rupture than the right. The explanation could be that passive venous congestion develops more readily in the left broad ligament because of the 90° angle of entrance of the left ovarian vein into the left renal vein. The dextrorotation of the uterus which occurs in up to 80% of cases may accentuate this predisposition [34].

But Mr. Misra, et al. [35] argued that the unscarred and spontaneous uterus ruptures frequently occur at the lower segment (weakest part), if the rupture part is the fundus, prior to the onset of labour. And the diagnosis is often delayed, because the haemorrhage is not revealed immediately, as blood collects in the intra-peritoneal cavity. Other studies also found that the rupture could occur at the lower uterine segment with higher frequency [10, 11], probably caused by the reduced thickness
which was due to the distention and elongation of the muscle fibers [26]. Ofir K ES, et al. found that cervical involvement was significantly more prevalent in the rupture of an unscarred uterus [11].

In our literature review, the fundus was found in 12.5% cases (1/8). The ruptures of posterior uterine wall and uterine anterior wall were found in 25% (2/8) and 12.5% (1/8) respectively. The rupture of lower segment was found in 12.5% cases (1/8). The rupture of right utero sacral area occurred in 12.5% (1/8) of the cases. The rupture places in the other two cases are unclear. However, the available data are insufficient to draw a meaningful conclusion on the most probable place where primigravida metrorrhexis without obvious inducement might occur.

Management

The basic treatment for a patient with a ruptured uterus is immediate resuscitation and surgery. The unscarred uterus rupture tends to be longitudinal and complete, hysterectomy is advised. However, for young females especially those who do not have children, it is better to preserve the uterus. Recent studies have shown tubal ligation shall not be carried out while double sea repair on uterus crevasse are recommended for females who want to preserve fertility. And the risk of the recrudescent metrorrhexis during gestation is between 4.3%-19% [36]. Martin [37] reported a rupture occurred at the lateral bottom of a uterus with a 19-week pregnancy and was treated by Gore-Tex soft tissue. Selective cesarean delivery was then performed in the 33th week of pregnancy and the repairing area was found intact and epithelized. It could be demonstrated that future pregnancies are allowed if the area could be repaired. For those who desire to continue to give birth caesarean section should be compulsorily selected in future gestation.

But in some cases, the patients often have severe tears not suitable for repair. In general, the surgical option must be individualized and should be dependent upon the type, location and extent of the rupture, as well as on the patient’s parity, the degree of bleeding, the available resources and desire to preserve her child bearing capacity [10]. If possible, repair is probably the best approach. In cases with wide bruises and contaminations, intractable uterine bleeding and multiple uterine rupture sites, longitudinal or low lying hysterectomy is preferable [1].

In our series, all the patients have received the repairmen of uterine crevasse. Four of them were treated by both repairment and bilateral tubal ligation. Four other cases lacked description for the management of fallopian tube. Ligation of the hypogastric artery could be useful for attaining adequate hemostasis [38].

Conclusions

The present case and review highlighted a few important lessons for medical practitioners. Although spontaneous uterine rupture in primigravid is extremely rare, there is still a possibility of occurrence. An unscarred uterus should not be considered “immune” to spontaneous uterine rupture. However, it is important to highly suspect for uterine rupture in primigravid women since it occurs often without warning, even in the absence of classical risk factors. Only through correct management, can optimal outcome be achieved and further complications be avoided.

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Disclosure of conflict of interest

None.

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SUUR in primigravida: case report and literature review


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