

Uterine rupture in pregnancy: two case reports and review of literature

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Summary

Rupture of a gravid uterus is an obstetric emergency. Risks factors include a scarred uterus but also spontaneous rupture of an unscarred uterus during pregnancy is possible. The authors present two cases of a spontaneous complete uterine rupture during pregnancy. The first case had only a past history of dilatation and curettage for abortion; the second case had a past history of dilatation and curettage for abortion and a monolateral laparoscopic salpingectomy for ectopic pregnancy. They presented with abdominal pain and after ultrasound scan, uterine ruptures were diagnosed. These cases show that there should be a high index of suspicion of uterine rupture in a gravid woman with a history of curettage for the possible presence of misunderstood uterine scar and in women with a past history of salpingectomy with or without corneal resection. Appropriate counseling and close follow-up might help to avoid such obstetrical catastrophes. To provide more insight into the possible risk factors for prelabor uterine rupture in pregnancy, a literature review was performed.

Key words: Uterine rupture; Dilatation and curettage; Uterine scar.

Introduction

Rupture of a gravid uterus is a rare surgical and obstetric emergency. The most accredited definition is from Plauche *et al.*: “those cases of complete separation of the wall of the pregnant uterus with or without expulsion of the fetus” [1]. Uterine rupture is more prevalent in less developed than in developed countries [2]. Risks factors include previous uterine surgery such as cesarean section, laparotomic or laparoscopic myomectomy, hysteroscopic surgery, iatrogenic uterine perforation, multiparity, previous instrumental abortion, inappropriate augmentation of labor, application of fundal pressure, placenta accreta, trauma, and congenital uterine anomalies [2-5]. It is possible to classify uterine rupture according to etiology: a) spontaneous rupture of previous scar (cesarean section, myomectomy etc); b) traumatic rupture of previous scar (version in obstetrics, accident, etc); c) spontaneous rupture of unscarred uterus; d) traumatic rupture of unscarred uterus.

Symptoms of uterine rupture are severe abdominal pain of sudden onset, palpable fetal body parts, cessation of contractions, signs of intraperitoneal bleeding, and all the features correlated to the hemorrhage that could lead to maternal and fetal (fetal distress, bradycardia) deterioration of vital signs leading up to shock. Less common symptoms are epigastric pain, shoulder pain (right-sided or bilateral), abdominal distension and paralytic ileus, ematuria, hypertonic uterus, altered uterine contour, and

fluid thrill. Often there is minimal external bleeding but an important internal bleeding with blood in the broad ligament and extaperitoneal spaces could be detected with an ultrasound examination. The typical ultrasound manifestations of uterine rupture are the empty uterus and the gestational sac above the uterus. Other sonographic findings are intrauterine blood and large uterine mass with gas bubbles [6]. Hruska *et al.* reported the importance of the MRI examination for assessment of pregnant patients in case of uterine rupture [7]. Treatment of uterine rupture is an early surgical intervention and previous hemodynamic stabilization of the patient where possible. It is necessary to correct hypovolemia after securing airway and oxygen administration. Maternal mortality is 0.44% and it resulted from hemorrhage, shock, sepsis, disseminated intravascular coagulation, pulmonary embolism, ileus paralyticus, peritonitis, and renal failure. It is possible to reduce fetal and maternal mortality with a prompt intervention, less than 18 minutes from onset of prolonged deceleration to delivery [8]. The authors present two cases of a spontaneous complete uterine rupture at a gestational age of 27 weeks in a 29-year-old patient and 34 weeks in a 38-year-old patient after previous misunderstood perforation. The cases were managed at the University of Cagliari (Hospital San Giovanni di Dio, Cagliari).

The first case had a past history of dilatation and curettage for abortion. The second case had a past history of di-

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lation and curettage for abortion and a monolateral salpingectomy laparoscopy for ectopic pregnancy. They presented with abdominal pain of sudden onset. After ultrasound scan, uterine rupture was diagnosed and an emergency laparotomy was performed and the following evaluation of the medical history gave a strong suspicion of a misunderstood perforation in the previous D&C. To provide more insight into the possible risk factors for prelabor uterine rupture in gravid, a literature review was performed.

Case Report

Case 1

The patient was 29-years-old, secondigravida, nulliparous with a gestational age of 27 weeks. She had previous appendectomy surgery and a dilatation and curettage for abortion 12 year prior. She reported a regular course of pregnancy until the moment when she arrived at the authors' Department for intense pelvic pain associated with a reduction in blood pressure (BP: 70/40). On physical examination a contracted uterus was appreciated and the ultrasound evaluation showed a placental detachment and free fluid in the peritoneal cavity; a fetal heart beat was present. An emergency laparotomy was performed for cesarean section to remove the fetus and placenta due to the occurrence of hemodynamic instability with anemia (hemoglobin 6 g/dl) and hypovolemic shock. After removal of the fetus and placenta and suction of 2,000 cc of hemoperitoneum, the uterus had a three-cm tear at the level of the left side wall of the uterine body; the authors performed a suture in double-layer with Vicryl n°1 in the breach of the uterine section and in the left side wall tear. During surgery 500 ml of whole blood was transfused to the patient, and another 500 ml was transfused after surgery. A male infant was delivered (Apgar 2-4; weight 950 grams) and the patient had an uneventful recovery. The child currently presents mild neurological deficits to a lower limb.

Case 2

The patient was a 38-year-old woman, gravida four, para one with a gestational age of 34 weeks. She had had in 2006 a dilatation and curettage for abortion and in 2007 a left salpingectomy laparoscopy for ectopic pregnancy. The patient presented at the authors' Department for pelvic pain and the uterus appear contract. A transabdominal ultrasound evaluation showed the presence of a corpuscular flap on the left hypocondrium with a 30-mm maximum thickness and a corpuscular hepato renal ground water with a 25-mm maximum thickness. Considering the subsequent hemodynamic instability of the patient (hemoglobin values from 8.7 g/dl to 6.6 g/dl) but a reassuring pattern at fetal monitoring, emergency cesarean delivery under general anesthesia was performed. After removal of the fetus and placenta and suction of 2,000 cc of haemoperitoneum, a seven-cm transverse full-thickness myometrial tear was observed at the bottom of the uterus; the authors performed a suture in double layer with Vicryl n°1. During surgery two units of blood and another unit after surgery were transfused. A male infant was delivered (Apgar 3-7; weight 2,530 grams) and the patient had an uneventful recovery. The child currently does not present any problems.

Discussion

Uterine rupture in an unscarred uterus

Uterine rupture in an unscarred uterus is a rarely and unpredictable obstetric complication. Real incidence of rup-

tured uterus is difficult to determine because of unknown causes of maternal deaths due to obstetric shock. The incidence of rupture of the unscarred uterus is inferior to one in 10,000 pregnancies in the most-developed countries [9]. Rachagan *et al.* reported a incidence that varied from 0.3/1000 to 7/1000 [10]. Sun *et al.* reported a rate of 0.012% [11].

The most important risk factors for unscarred uterine rupture are multiparity, the inappropriate use of oxytocin, mid-forceps delivery, uterine over-distention due to the presence of twin pregnancy, previous curettage, and congenital anomalies. The most common sign in women with a uterine rupture without a scar is shock, followed by uterine bleeding, severe abdominal pain, and easily palpable fetal parts. Traditionally, primigravidae and unscarred uteri are considered immune to rupture, but the present authors found in literature some cases of nulliparous with a previous history of diagnostic hysteroscopy and uterine perforation at 35 weeks [12]. The past history of curettage, diagnostic or operative hysteroscopy, can suggest an unknown uterine perforation [13]. Multiparity is a important risk factor and many cases in literature are described [14, 15]. Tarney *et al.* described a case of uterine rupture in third trimester of an unscarred uterus in a quadruplet pregnancy in a 30-year-old woman that had not undergone previous surgeries; a high-order gestations may be an independent risk factor for uterine rupture [16]. Mamour *et al.* described a case of spontaneous uterine rupture during pregnancy at 35 weeks of an unscarred uterus before labour; her obstetrical history revealed a multipara patient with a history of four pregnancies that ended spontaneously by vaginal delivery. In this case high parity was recognized as major risk factor of spontaneous uterine rupture in unscarred uterus. A such case described Sun *et al.* where high parity was the only risk factor in a patient who presented upper abdominal discomfort and vomiting for three days; during emergency laparotomy, the entire amniotic sac was found in the peritoneal cavity with a rupture of the uterine fundus. The literature describes cases where multiparity is the main risk factor [17]. Another risk factor is an abnormal adherence of placenta to the uterine wall without interposition of deciduas basalis. Placenta percreta is the rarest form but may complicate the pregnancy with acute severe hemorrhage [18]. Pierzynsky *et al.* reported a case of uterine rupture due to placenta percreta in otherwise uncomplicated pregnancy; a 35-year-old, gravid 5, para 5, at 15 weeks two days gestation with negative history of uterine scarring developed symptoms of incipient hypovolemic shock and on exploratory laparotomy found a midline uterine rupture infiltrated by the placenta. Abnormal placentation should be taken into consideration also in women in the second trimester who have no history of uterine instrumentation [19].

Also, Xia *et al.* reported a case of uterine rupture that was associated with placenta accreta with a spontaneous uterine perforation in the second trimester of pregnancy with mul-

multiple perforations of an unscarred uterus revealed by hemoperitoneum at 22 weeks. Emergency total hysterectomy was performed [20]. Another risk factor is the use of prostaglandins and oxytocin to induce labor [21, 22]. Cuelar Torriente presented a case of silent uterine rupture in an unscarred uterus during pregnancy at third trimester abortion by use of mifepristone and misoprostol in a patient who had a history of intrauterine procedures. They suggested that this uterine rupture resulted from an unrecognized perforation in a previous intrauterine manipulation. Many cases have been reported of uterine rupture in an unscarred uterus during second trimester pregnancy termination with intravaginal misoprostol [23]. Chang *et al.* reported a case of a multiparous who had a previous vaginal birth delivery and who was labor-induced with PGE2 and oxytocin. Even if one of the most risk factors are incorrect administration of oxytocin in multiparous woman, in this case the authors excluded the possibility to an augmentation of labor and stressed on the possibility of a silent rupture due to a previous pregnancy [24]. Also, adenomyosis can be a risk factor for uterine rupture due to the weakening of the uterine muscle fibers; in a case report with review of literature, Nikolaou *et al.* reported a rare case of spontaneous uterine rupture of an unscarred uterus caused by adenomyosis in the early third trimester [25-27]. Agarwal *et al.* reported a case of intrapartum unscarred uterine fundal rupture in a case of drug abuse. A careful history of drug abuse must be elicited when the common causes of uterine rupture have been excluded or the rupture site is unusual. There are other described cases of uterine rupture associated with cocaine abuse [28, 29].

Also, the cause of uterine rupture could be uterine diverticulum, frequently misunderstood and reported as uterine sacculation. Uterine diverticulum has a narrow connection with the uterine cavity and a thicker wall than in sacculation. While uterine sacculation is usually observed during pregnancy, diverticulum is usually detected in non-pregnant women. Uterine diverticula as a complication during pregnancy are rare. Rajah *et al.* reported a primigravida woman in whom an MRI revealed uterine diverticulum in the 22 weeks of gestation. A cesarean section was performed in the 31st week. The diverticulum originated from the posterolateral wall of the uterine body and did not contain the fetus. The diverticulum was not excised due to surgical risks. The authors considered that the underlying etiology for the diverticulum may have been congenital because this patient was primigravida with no prior cervical or uterine procedure [30]. Matsubara *et al.* described a case of a primigravida woman with a thin anterior uterine wall, a feature compatible with incomplete uterine rupture; her condition was detected by abdominal palpation and ultrasound. This case suggest that an unscarred primigravida pre-labor uterus can show the features of incomplete rupture even in the absence of discernable risk factors and that abdominal palpation and ultrasound may be useful in diagnosis. They suspected a rupture uterine diverticulum [31].

In literature there are six cases with uterine rupture in case of uterus congenital anomalies; five are related to the presence of bicornuate uterus, and only one relates to septate uterus. Damiani reported the first case of uterine rupture in a septate uterus; the authors assumed that it could lead to uterine overdistension due to the presence of the medial sept [32], but also to the history of a D&C which could have caused an unknown perforation or a weakness of uterine wall [33]. Often a lack in anamnesis can miss important risk factors for uterine rupture.

Uterine rupture in scarred uterus

Due to an improvement rate of cesarean births, several large studies have identified the risks of maternal morbidities associated with a trial of labor after cesarean delivery. Uterine rupture during labor is a serious and uncommon obstetrical complication that can lead to severe prognosis for the mother and her child if not immediately diagnosed and treated. Rupture of the uterus in labor is also associated with cessation of labor pain, recession of presenting fetal body parts, cervical lacerations, and vaginally palpable uterine defect. One series reported that 81% of patients with uterine rupture during labor have evidenced fetal distress prior to the onset of bleeding or abdominal pain [34, 35]. The American College of Obstetricians and gynecologists (ACOG) suggested that a trial of labor among patients with a previous cesarean section is reasonable. The percentage of uterine rupture in case of trial of labor is reported to range from 0.5% to 1%, is associated with cessation of labor pain, recession of presenting fetal body parts, cervical lacerations, vaginally palpable uterine defect, fetal distress. Rodriguez *et al.* reported that 81% of patients with uterine rupture during labor have evidence of fetal distress prior to the onset of bleeding or abdominal pain [35]. Ho *et al.* reported a case of uterine and bladder rupture during the second stage of labor in a 39-year-old patient with a previous cesarean section [36]. The risk of uterine rupture is higher in patients undergoing induction of labor after previous cesarean section when compared with that in spontaneous labor. Lin *et al.* reported a higher rate of rupture in patients induced with misoprostol and oxytocin, and suggested that the risk of rupture was higher in patients with multiple prior cesarean section [37]. Even if the American College of Obstetricians and Gynecologists Practice Patterns Committee concluded the oxytocin was not contraindicated for induction and augmentation of labour; based on the current literature, the use of oxytocin increase the risk of uterine rupture [38-40]. Grobman *et al.* reported that if the woman has an anamnesis of one prior vaginal delivery and one prior cesarean section, the risk of uterine rupture in labor is similar to the normal population [41].

Bujold *et al.* analyzed the cervical status before labor induction [42]. Although not statistically significant, the au-

thors found a trend towards a higher rate of uterine rupture in patients with a lower modified Bishop's score before labor induction. Harper *et al.* reported an increase of the risk in women with a cervical exam of < 2 cm and 2 to 3.9 cm at the initiation of oxytocin: "Women who received oxytocin starting at > 4 cm had a similar risk of uterine rupture as women who labored spontaneously" [43]. It seems to be important to consider the maximum dose of oxytocin for induction. Cahill *et al.* suggested that an upper limit of 20 mU/minute seems related to a reasonable risk of uterine rupture (1%) compared with upper doses ranging from 20 and 30 mU/minute, which are related to a risk of uterine rupture that ranged from 2.9% and 3.6% [44].

Another risk factor is the number of previous cesarean deliveries, as reported by Miller *et al.* that found a percentage of 1.7% of uterine rupture in patients with more than two cesarean sections, compared with a 0.6 % in case of single previous cesarean section [45]. Stamilio *et al.* reported a higher risk of uterine rupture if the interpregnancy interval was less than six months, with a percentage of uterine rupture of 2.7% [46]. A surgical risk factor is the type of suturing. The single-layer locked continuous suturing was associated with a higher uterine rupture risk than double layer closure [47]. Cahill *et al.* reported that women with twin gestations are less likely to attempt a VBAC, but they are not more likely to fail a VBAC trial or experience a major morbid event compared with women with singleton gestations [48]. Even though uterine rupture is a rare complication the consequences are of great clinical significance. The choice of a VBAC should be discussed with the patient because it could be important in their decision-making process. Uterine rupture may also be associated to oral misoprostol administration for induction of labor for termination of pregnancy. Chapman *et al.* found the incidence of uterine rupture was significantly higher among women with a prior cesarean section (three years 3.8% vs. one year 0.2%). All four uterine ruptures were in pregnancies of 22-24 weeks' gestation and had oxytocic induction agents [49].

Lu *et al.* reported an unusual case of uterine rupture caused by intra-amniotic ethacridine used for second trimester pregnancy termination with the expulsion of the fetus into the broad ligament through the lateral wall [50]. In contrast to second and third trimester uterine rupture, uterine rupture during the first trimester is an extremely rare complication, but as well life threatening cause of intraperitoneal hemorrhage. Some cases of a spontaneous first trimester rupture of the pregnant uterus following a previous cesarean section are described in literature [51]. Often first trimester uterine ruptures are related to scar dehiscences after previous cesarean sections [52]. Sliutz *et al.* postulated their hypotheses that all first trimester uterine ruptures are caused by scar implantation of the trophoblast [53].

Conclusions

Rupture of the non-labouring uterus is rare event in which the life of both the mother and the child are in danger. Spontaneous ruptures are almost always intrapartum and risk factors that can predispose to uterine rupture are a cesarean section scar, advanced maternal age, uterine abnormalities, grand multiparity, macrosomic fetus, cephalopelvic disproportion, and uterine trauma from prior instrumentation from abortion, version and oxytocin stimulation [54-60]; also risk factors associated with early uterine rupture in pregnancy include iatrogenic uterine perforation during hysteroscopy procedures, previous salpingectomy, and corneal resection following ectopic pregnancy and myomectomy laparoscopy or laparotomy [61, 62]. Other less common causes are placenta increta, congenital anomalies, trauma, and sacculation of entrapped retroverted uterus. Rupture of a cesarean section scar is usually considered to be the most common predisposing factor [63]. Rupture in a uterus without previous surgery is rare and most often associated with grand multiparity and long, obstructed labour, whereas traumatic rupture is often secondary to mechanical intervention, such as forceps delivery. The incidence ranges from one in 8,000 to one in 15,000 deliveries [64, 65]. It has been suggested that predisposing risk factors to such unexpected uterine rupture may include uterine diverticulae [66], arteriovenous malformation, and injudicious oxytocic stimulation [67]. Uterine rupture can also occur without these factors. Manouana *et al.* reported a case of uterine rupture during labour in which no predisposing factor existed, be it medical, surgical, gynecological or obstetrical [68]. In our cases, the patients had previous episodes of uterine curettage for missed abortion. We think that their patients may have had uterine scar from a unnoticed incomplete or complete uterine perforation during curettage. The perforation may have been situated at the uterine fundus and subsequently the products of conception of the current pregnancy may have embedded in that region, so that following continuous distension of the uterus with evolving pregnancy, there was a stretch of the weakened area leading finally to rupture and protrusion of the amniotic sac with the placenta into the peritoneal cavity. This hypothesis has already been made by Bevan *et al.* [69]. In literature there are few described cases of uterine rupture in pregnancy preceded by a dilatation and curettage for abortion because the lesion uterine remains most often misunderstood [70-72]. Also, in the latter case, the patient in the past had underwent a monolateral salpingectomy for ectopic pregnancy; it is believed that salpingectomy with corneal resection attenuates uterine musculature at the corneal region which can lead to a subsequent rupture of the uterus in the early course of pregnancy. The presence in history of previous minor surgery, such as curettage, should be considered a possible risk factor for uterine rupture during labor but also in the second trimester, resulting in a greater focus on the patient's symptoms during pregnancy and the

delivery. The postoperative course of previous interventions should investigate important symptoms such as fever or delayed discharge.

The presented cases show that there should be a high index of suspicious of uterine rupture in a gravid woman with a history of curettage for the possible presence of uterine scar misunderstood or for the presence of uterine scar understood and in women with a past history of salpingectomy with or without corneal resection.

Uterine rupture should be considered in the differential diagnosis in all pregnant patients who present with acute abdomen, show fluid collection in the peritoneal cavity, and have specific risks factors.

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