



Uterine rupture in pregnancy after an intervention complicated by uterine perforation: Case report and systematic review of literature

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ABSTRACT

Objectives: The present study is a descriptive study of characteristics of women who had a uterine rupture during pregnancy with a history of uterine perforation and no previous caesarean section.

Study design: We present a case report of a woman with a uterine rupture in pregnancy subsequent to a perforation made by uterine sounding and we performed a systematic review including all case-reports of uterine ruptures after perforation during dilatation and curettage or due to uterine sounding.

Results: 14 case-reports were included in this review. 12 out of 14 women presented with abdominal pain prior to the uterine rupture. In eight out of 14 cases an abdominal ultrasound was performed and in five ultrasounds a uterine wall defect was detected, in two other cases free fluid was visible and in one case fetal bradycardia was seen. Neonatal outcome was uneventful in six cases, there where two immature fetuses born and in two cases there was fetal demise.

Conclusion: Uterine rupture in a (supposed) unscarred uterus is a relatively unknown complication. We recommend clinicians to be aware of uterine rupture in pregnant women with abdominal pain and a history of uterine manipulation. When a uterine rupture is suspected and mother and fetus are in suspected good condition, an ultrasound examination could be an easy and fast next step.

Introduction

Uterine rupture is a rare event in pregnancy and significantly increases the risk for maternal and fetal morbidity and mortality [1]. The most important risk factor for a uterine rupture is the scarred uterus [2]. Incidence of uterine rupture during labour in women with a previous caesarean section ranges from 325 to 468 per 100,000 women undergoing trial of labor [3,4]. Less data on uterine ruptures during pregnancy or labour is available for women with no history of caesarean section who had a uterine perforation made by dilatation and curettage or uterine sounding [5]. We report a case of a uterine rupture in the third trimester of pregnancy after a fundal perforation made a year before, and performed a systematic review of the literature.

Case report

A healthy 28-year-old female (gravida 7, parity 1), pregnant with a dichorionic twin, was admitted to the hospital at 34 weeks and 6 days of gestation with uterine contractions and a progressive pain in the left upper abdomen.

The current pregnancy was a result of intracytoplasmic sperm injection (ICSI). After her first ICSI cycle, she underwent 6 attempts of embryo replacement since December 2014 with unsuccessful outcome. For the second ICSI cycle she went to Cyprus where 23 out of 26 oocytes got fertilized. One out of 3 eligible embryos was transferred into the uterus which resulted in an ongoing dichorionic twin pregnancy. The first trimester was complicated by ovarian hyperstimulation syndrome. Two torsions of the right ovary occurred, at a gestational age of 9 and 14 weeks, for which she underwent two successful laparoscopic detorsions of the ovary. During the first and second trimester she was admitted several times for hyperemesis gravidarum and vaginal bleedings. The vaginal bleedings were of unknown origin, PAP smears were normal, there was no infection or low-lying placenta. During the last four weeks of pregnancy, multiple ultrasounds were performed, because of suspected fetal growth retardation and decreased fetal movements of both fetuses.

Obstetric history consisted of recurrent miscarriages, for which no cause was found. She had one live birth in 2013 at a gestational age of 36 weeks and 5 days. Labour was induced due to suspected fetal growth retardation and decreased fetal movements. In the second stage of

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Fig. 1. The abdominal ultrasound shows a. the place of perforation made prior to uterine rupture and b. the membranes partially expelled through this defect.

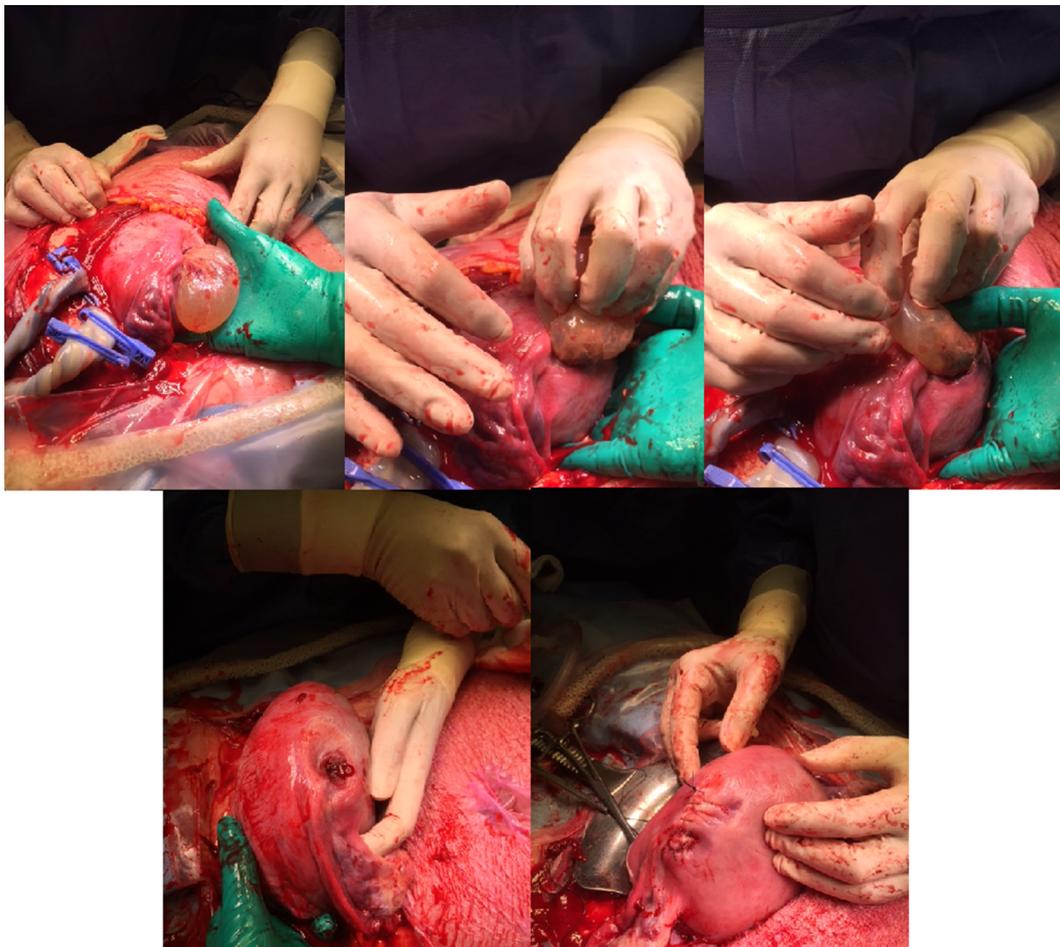


Fig. 2. The uterus was lifted outside the abdominal cavity after the delivery. It shows us a + b + c. the membranes partially expelled through the defect. d + e the defect after closing it in two layers.

labour, an instrumental vaginal delivery was indicated because of fetal distress. A girl was born with a birthweight of 2240 g ($p = 20\text{--}50$) and an Apgar score of 7 at 1 min and 8 at 5 min.

Her gynaecological history consists of a diagnostic laparoscopy in August 2015. The procedure was performed because of persistent complaints of a stinging and cramping sensation in the lower quadrant of the abdomen that increased in frequency and was not related to the menstrual cycle. These complaints were present for at least 5 years and no explanation was found, neither on vaginal examination nor transvaginal ultrasound.

Prior to the laparoscopic procedure the uterus was measured with a sound and a Valchev uterine manipulator was inserted. On following

laparoscopy a uterine defect was visualized and it was recognized as an accidental perforation, caused by the sound. The perforation was coagulated (Fig. 1A). There were no pelvic abnormalities otherwise. The adnexa were both normal, there was slightly notable vascularisation of the Douglas cavity and no signs of endometriosis.

During the current admission she had no complaints other than uterine contractions and a progressive pain in the left upper abdomen. There was no vaginal bleeding or amniotic fluid loss, and there were normal movements of both fetuses. Cardiotocogram showed two reassuring fetal heart rate tracings and 3 uterine contractions every 10 min. Vaginal examination showed a mediosacral, effaced and weak cervix, 4 cm of dilatation, and a cephalic position of fetus A at Hodge 1.

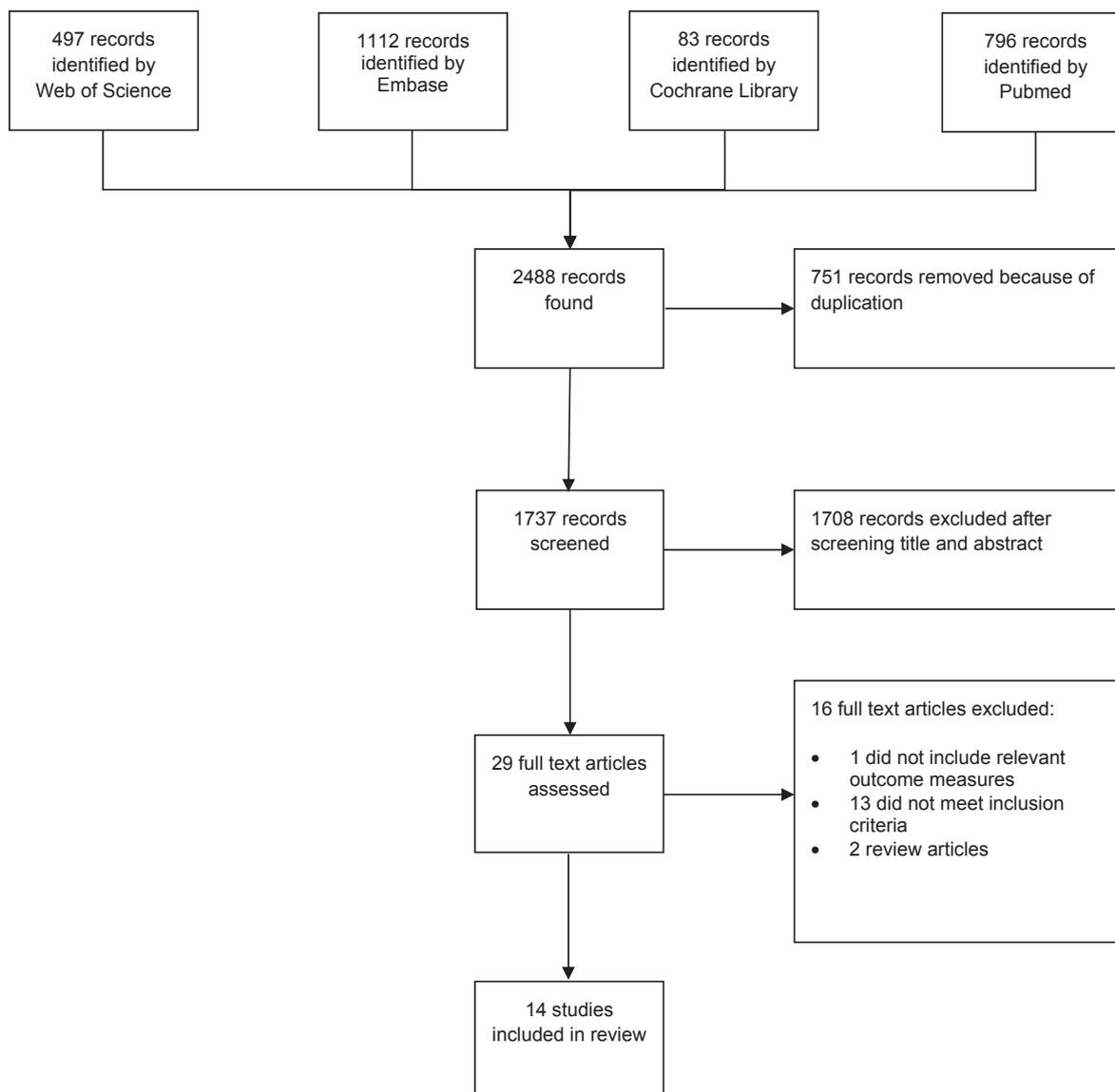


Fig. 3. Flow chart. 13 studies were included in this review and our own case report was added which makes it a total of 14 studies included in this review.

The pain in the left upper abdomen was progressive and more prominent during uterine contractions. Ultrasound examination showed both fetuses in cephalic position with normal amniotic fluid. At the indicated painful region in the upper left abdomen, a uterine wall defect of 2 by 4 cm was shown. Membranes were partially expelled through the defect in the abdominal cavity (Fig. 1B).

To prevent the mother and fetuses from further damage, a Caesarean delivery was performed. Manual examination of the uterus from inside the abdomen before delivery of the twins confirmed a uterine wall defect and presentation of membranes filled with amniotic fluid through the defect into the abdominal cavity. Two boys were born with an Apgar score of respectively 8/6/8 and 8/8/9, and birth weights of 2299 g (p20-50) and 2100 g (p16-p20). Umbilical arterial pH was not performed. The neonates were admitted to the neonatal medium care unit because of prematurity. After delivery of the fetuses the uterus was lifted outside the abdominal cavity (Fig. 2).

The location of the defect on the left side of the fundus corresponded with the location of the perforation made during laparoscopy a year before (Fig. 2A). The defect in the fundus was closed in two layers (firstly myometrium and afterwards serosa) with Vicryl 0 (Fig. 2E). Total blood loss was 250 ml. Two weeks after delivery, patient and neonates were discharged in good condition.

Material and methods

In this review, we included all case reports of women with a uterine rupture in pregnancy subsequent to a previous perforation due to dilatation and curettage or due to uterine sounding.

Exclusion criteria were women with previous caesarean section and/or uterine surgery that includes deliberate scarring of the myometrium and/or opening of the endometrial cavity.

We searched the Cochrane Library, Pubmed, Embase, and Web of Science (till February 22th 2017). The following terms were used: (“Hysteroscopy”[Mesh] OR “Laparoscopy” [Mesh] OR “Salpingectomy”[Mesh] OR “Salpingostomy”[Mesh] OR Hysteroscop*[tw] OR laparoscop*[tw] OR salpingectom*[tw] OR curettage *[tw] OR tubectom*[tw] OR salpingostom*[tw]) AND (“Uterine Rupture”[Mesh] OR ((Uterine[tw] OR uterus[tw]) AND ruptur*[tw])).

We retrieved all titles and abstracts identified by electronic searching. We removed duplicates and the remaining references were examined by two review authors (SH and ME) independently. These authors screened the titles and abstracts and eliminated articles that were obviously not relevant to the search question. When both authors (SH, ME) determined that the article might be eligible for inclusion, the

Table 1
Characteristics.

Characteristics	N %
Age (mean, 25–75th percentiles)	30,7 (28–33)
Prior surgery ^a	
Dilatation and curettage	8 (57%)
Laparoscopy	6 (43%)
Hysteroscopy	2 (14%)
Treatment of perforation	
Electrosurgery	1 (7%)
Sutured	1 (7%)
No treatment	8 (57%)
Unknown	4 (29%)
Time between perforation and pregnancy, (years, 25–75th percentiles)	5 (3–6)
Mode of conception	
Natural	1 (7%)
Fertility treatment	4 (29%)
Unknown	9 (64%)
Pregnancy	
Singleton	9 (64%)
Multiple	3 (21%)
Unknown	2 (14%)
Trimester	
First	1 (7%)
Second	1 (7%)
Third	10 (70%)
unknown	2 (14%)
Complaints ^a	
Abdominal pain	12 (86%)
Vaginal bleeding	1 (7%)
Acute abdomen	5 (36%)
No complaints	2 (14%)
Mode of diagnosis	
Abdominal ultrasound performed	8 (57%)
Rupture found on ultrasound	5 (63%)
Perioperative hemorrhage (range)	0–3500 ml
Treatment for rupture	
Sutured	9 (64%)
Hysterectomy	2 (14%)
No treatment	1 (7%)
Unknown	2 (14%)
Outcome mother	
Uneventful	13 (93%)
Unknown	1 (7%)
Outcome fetus ^b	
Uneventful	7 (50%)
Immature fetus	2 (14%)
Fetal demise	2 (14%)
Cerebral palsy	1 (7%)
Unknown	2 (14%)
Subsequent pregnancies	
Uneventful	1 (7%)
Unknown	13 (93%)

^a Some women had more prior surgeries or complaints causing the total sum extend 100%

^b Three case-reports describe a twin pregnancy of which two cases had good fetal outcome for both fetus, one case had one fetal demise and one uneventful outcome (in the table placed under ‘fetal demise’)

full text article was obtained. Each author independently determined whether those trials were eligible for inclusion. Any disagreements were resolved by discussion. The data were extracted using pre-specified extraction sheets made for this review. Data were checked for accuracy and entered into a table. When information was insufficient or unclear, authors of the original articles were contacted to provide further information. Descriptive variables were collected for age, type of prior surgery (dilatation and curettage, laparoscopy or hysteroscopy, treatment for perforation (electrosurgery or sutured), time between uterine perforation and pregnancy, mode of conception (natural versus fertility treatment), type of pregnancy (singleton or multiple), trimester at time of uterine rupture, complaints at time of uterine rupture in pregnancy, mode of diagnosis of uterine rupture, perioperative

haemorrhage, treatment for uterine rupture, maternal and neonatal morbidity and mortality, and potential lag time for a subsequent pregnancy after the event.

Results

We identified 2488 articles, 796 in Pubmed, 1112 in Embase, 497 in Web of Science and 83 in the Cochrane Library. After removal of duplicates we assessed 1737 articles on title and abstract, of which 1708 were excluded. Of the remaining 29 articles that were assessed full text, another 16 were excluded: 1 did not include relevant outcome measures, 13 did not meet inclusion criteria and 2 were review articles. Therefore, 13 case-reports were left to be included in this review, to which we added our own case report (Fig. 3). Of these 14 case reports, four did not document on a perforation prior to pregnancy, but as these women did undergo either dilatation and curettage or uterine sounding, it is possible that a perforation occurred and remained unrecognized at the time of surgery. Therefore, these cases were included as well (Table 1) [6–17]. (see. Table 2)

Maternal age ranged from 23 to 41 with a median of 30.7 years (Table 1). Of the perforations made during dilatation and curettage or by uterine sounding, two were electro coagulated, one was sutured and eight were left untreated (Table 1). Time between the perforation and pregnancy ranged between zero and 12 years. 12 out of 14 women presented with abdominal pain. In one case the abdominal pain existed 20 days postpartum before the uterine rupture was discovered. One case presented with abdominal pain and vaginal bleeding. In five cases an acute abdomen is described as clinical presentation of the uterine rupture. In two cases uterine rupture was an incidental finding and there were no complaints (Table 1).

In eight out of 14 cases an abdominal ultrasound was performed, and in five the uterine wall defect was noted. In the three other cases fetal bradycardia and free intra-abdominal fluid was seen. Eight emergency caesarean sections were performed, one due to maternal shock, one due to vaginal bleeding at 8 cm of dilatation and six because of findings on the abdominal ultrasound. In one case an elective caesarean section was planned because of failed induction without signs of a uterine rupture. (Table 1).

In 13 cases the recovery of the mother was uneventful, one did not describe maternal outcome (Table 1). Fetal outcome was reported as good in six cases. In one case an infant was delivered with an Apgar score of 2 and 4 at respectively after one and five minutes. The child currently presents mild neurological deficits to a lower limb. Another case reported a twin pregnancy with fetal demise of one of the two. Two of the ruptures occurred in first and second trimester, too early for the fetus to be viable. One uterine rupture was diagnosed 20 days postpartum, after an uncomplicated delivery. The patient came to the hospital with persistent lower abdominal pain and intermittent fever, pelvic sonography showed a low segment anterior wall defect of the uterus. In two cases the clinical outcome is not described (Table 1).

Comment

To our knowledge, this is the first systematic review on uterine rupture during pregnancy in women with a history of perforation due to uterine sounding or dilatation and curettage. Uterine rupture in a (supposed) unscarred uterus is a relatively unknown complication and therefore important to gain awareness among health care providers.

Different methods were used dealing with the perforation. Current literature does not answer the question whether and how a uterine perforation should be repaired, A theory put forward by Al-Kufaishi et al. is that subsequent poor vascularisation at the perforation site after coagulation may cause less favorable healing circumstances. The defect may therefore be more predisposed for forcing influences and rupture [11]. Perhaps suturing the defect or no treatment when bleeding is not significant would decrease the possibility for uterine rupture in future

Table 2
Main information of included studies.

Article	Age	Prior surgery	Treatment of perforation	mode of Conception
Ajuriagoaskoa	33	Curettage + hysteroscopy	Unknown	Unknown
Al-Kufashi	29	Laparoscopy	Sutured	IVF
Dow	34	Laparoscopy + dilatation and curettage	No treatment	Unknown
Fedorkow (1)	Unknown	Dilatation and curettage	Unknown	Unknown
Fedorkow (2)	Unknown	Dilatation and curettage	Unknown	Unknown
Ficicioglu	29	Laparoscopy	No treatment	IVF
Heemskerk	28	Laparoscopy	No treatment	ICSI
Ijaz	32	Dilatation and curettage	No treatment	Unknown
Mannini	34	Dilatation and curettage	No treatment	Natural
Pontis	29	Dilatation and curettage	No treatment	Unknown
Reed	23	Dilatation and curettage	No treatment	Unknown
Sun	28	Laparoscopy	No treatment	Unknown
Taylor	28	Laparoscopy	Electrosurgery	Unknown
Uccella	41	Hysteroscopy	Unknown	IVF

Article	Pregnancy	Gestational age (weeks)	Complaints	Mode of diagnosis
Ajuriagoaskoa	Singleton	35 + 2	Acute abdomen	Unknown
Al-Kufashi	Singleton	33	Abdominal pain	Ultrasound
Dow	Singleton	34	Abdominal pain	Ultrasound
Fedorkow (1)	Unknown	Unknown	Abdominal pain	Unknown
Fedorkow (2)	Unknown	Unknown	No complaints	Unknown
Ficicioglu	Multiple	29	Abdominal pain + vaginal bleeding	Surgery
Heemskerk	Multiple	34 + 6	Abdominal pain	Ultrasound
Ijaz	Singleton	8	Acute abdomen	Ultrasound (free abdominal fluid)
Mannini	Singleton	15	Abdominal pain	Ultrasound
Pontis	Singleton	27	Acute abdomen	Ultrasound (free abdominal fluid)
Reed	Singleton	42	No complaints	Surgery
Sun	Singleton	38	Abdominal pain	Ultrasound
Taylor	Singleton	35	Abdominal pain	Surgery
Uccella	Multiple	35	Acute abdomen	Ultrasound (fetal bradycardia)

Article	Treatment of rupture	Outcome mother	Outcome fetus	Subsequent pregnancy
Ajuriagoaskoa	Hysterectomy	Unknown	Uneventful	Unknown
Al-Kufashi	Sutured	Uneventful	Uneventful	Unknown
Dow	Sutured	Uneventful	Uneventful	Unknown
Fedorkow (1)	Unknown	Uneventful	Unknown	Unknown
Fedorkow (2)	Hysterectomy	Uneventful	Unknown	Unknown
Ficicioglu	Unknown	Uneventful	Uneventful	Unknown
Heemskerk	Sutured	Uneventful	Uneventful	Unknown
Ijaz	Sutured	Uneventful	Immature fetus	Unknown
Mannini	Sutured	Uneventful	Immature fetus	Unknown
Pontis	Sutured	Uneventful	Cerebral palsy	Unknown
Reed	No treatment	Uneventful	Uneventful	Uneventful
Sun	Sutured	Uneventful	Uneventful	Unknown
Taylor	Sutured	Uneventful	Fetal demise	Unknown
Uccella	Sutured	Uneventful	Child A: uneventful; Child B: fetal demise	Unknown

pregnancy.

In this review, 27% of all pregnancies following a perforation were established after fertility treatment. However, due to the small number of available cases in literature we can not state this as a possible risk factor. Another limitation of this study is the moderate follow-up of the different cases, making it hard to find possible other risk factors for uterine rupture after perforation.

Women with a history of caesarean section are advised to wait at least 12 months to one year before imitate a new pregnancy, to decrease the change of uterine rupture [18]. In this review the time between the perforation and uterine rupture varied from 0 months to 12 years. Therefore a recommendation regarding the time between a perforation and subsequent pregnancy is hard to make, however we would like to suggest a similar time span as women who underwent a caesarean section.

A variety of clinical manifestations is described when uterine rupture is present. A rupture of the uterus happens most often during labour since the force of the contractions weakens the uterine wall. Abdominal pain is a common complaint, but can be hard to differentiate from contractions. A continuous or progressive abdominal pain

should make one alert of a possible uterine rupture. A quick diagnosis and treatment is very important for maternal and fetal outcome. In eight case reports ultrasound was used, five showed the defect and three showed free fluid. This suggests that ultrasound in the diagnosis of uterine rupture could be a fast and accurate method.

In conclusion, we recommend that clinicians should be aware of the possibility of a uterine rupture in cases of acute abdominal pain during pregnancy, in women with an unscarred uterus, with a prior history of uterine manipulation, such as dilatation and curettage or sounding. If a rupture of the uterus is considered, and maternal and fetal condition is acceptable, an ultrasound is an easy and quick way to diagnose the defect.

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Appendix A. Supplementary material

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.srhc.2018.11.001>.

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