Recurrence of a second-trimester uterine rupture in the fundus distant from old scars: A case report and review of the literature

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A B S T R A C T

Uterine rupture is a rare event which can have severe obstetric consequences and most often occurs on a scarred uterus at the site of the scar. However, a uterine rupture can appear at another location. We report the case of a woman with a previous emergency caesarean section for spontaneous posterior uterine rupture which recurred at another site during her second pregnancy. She was admitted to the emergency room with acute abdominal pain and development of a pre-shock hemorrhagic state. Abdominal ultrasound showed abundant peritoneal fluid and a fetus without cardiac activity in an intact bulging amniotic membrane. We performed an emergency laparotomy, which confirmed an intact amniotic sac in the abdominal cavity and showed a 7 cm transverse fundal uterine rupture beginning at the right angle, distant from the old known scars. In view of the high maternal and fetal morbidity, obstetricians should have a high suspicion of an antepartum uterine rupture, even at an early gestational age, in the event of acute abdominal pain over a scarred uterus.

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1. Introduction

Spontaneous uterine rupture in the second trimester is a rare but life-threatening complication for both mother and fetus, particularly because of a delayed diagnosis. The majority of ruptures occur in the third trimester, during labor, following induction or after a previous caesarean section. Although rare, their incidence in a non-scarred uterus is increasing and is associated with higher maternal and fetal morbidity [1]. The literature on primary antepartum uterine rupture is limited to a few case reports. Generally, no direct etiology is found; however, risk factors have been described, such as acquired, congenital or iatrogenic myometrial weakness.

We present the rare case of recurrence of spontaneous rupture at 23 weeks of gestation with intrauterine fetal demise. The singularity of our case is the occurrence of a second spontaneous rupture located at a site other than the old scars (the posterior uterine rupture and caesarean section scar). We reviewed the recent literature regarding cases of spontaneous uterine rupture which did not occur during labor.

2. Case Presentation

A 29-year-old patient at 23 weeks of gestation of her second pregnancy was admitted to hospital for sudden onset of diffuse abdominal pain without evident triggering factors. Two years earlier, she had had an emergency caesarean section at 36 weeks of gestation due to a pathological fetal heart rate pattern leading to an intraoperative discovery of a posterior isthmic uterine rupture. She also reported a laparoscopic cystectomy for endometrioma.

On examination, her blood pressure was 112/65 mmHg and her heart rate was 90 beats per minute. The uterine fundus was painful to palpation. Repeated emergency ultrasound examinations revealed free intra-abdominal fluid with clots (Fig. 1) and the absence of fetal heartbeat. An emergency laparotomy through a transverse suprapubic incision was performed, which revealed the expulsion of the fetus within his amniotic sac in the abdominal cavity (Fig. 2a and b). A uterine rupture of 7 cm was present from the right uterine angle and extending towards the left angle (Fig. 3). The omentum and the right tubal pavilion were adherent to the posterior ruptured uterine serosa. Therefore, adhesiolysis was performed. During abdominal inspection, the round ligaments and the ovaries were found to be normal, although fine peri-ovarian adhesions were present. Manual revision of the uterine cavity revealed decidual fragments. After resection of the margins, we performed a hysterorrhaphy with a single layer of simple interrupted suture due to myometrial friability. During the procedure, the patient was given two units of red blood cells and two units of fresh frozen plasma.
The patient's post-operative course was uneventful, with full recovery. Eight weeks after surgery, the integrity of the fundal scar was inspected by hydrosonography (Fig. 4). Additionally, the patient received genetic counselling. There was no known history of collagen disease in the family. The patient was recommended laboratory testing for an Ehlers-Danlos syndrome Type IV or Loeys-Dietz syndrome but declined.

3. Discussion

Rupture of the pregnant uterus is a life-threatening emergency. In developed countries, the prevalence of uterine ruptures over caesarean section scarring is about 1% [2], while ruptures over non-scarred uterus are even rarer, occurring in 1/5700 to 1/20,000 pregnancies [3]. The main risk factors for spontaneous ruptures include: history of uterine surgery (caesarean section, myomectomy, salpingectomy, perforation during mobilization), short time interval (less than 6 months) between surgery and pregnancy or between 2 pregnancies, multiparity, advanced maternal age, multiple pregnancies and placental abnormalities. Other predisposing factors include Ehlers-Danlos syndrome, cocaine abuse, in utero exposure to diethylstilbestrol, uterine abnormalities, adenomyosis, labor dystocia and use of uterotonic drugs [1,4–6,20].

Among the seventeen cases of spontaneous uterine rupture during the second trimester reported in the literature, the fundus represents the most common site (Table 1). Among the nine fundal ruptures, there are two cases of invasive placentation, two cases of adenomyosis, one ruptured cornual pregnancy, one septated uterus and one previous laparoscopic examination. No risk factors were identified for the last case.

The second most common site is the posterior wall. In two out of three cases, the site corresponded to an old myomectomy scar; in one of these three cases, a curettage was noted in the history. Other sites corresponded to the horns, the anterior wall and the lower segment (Table 1). The peculiarity of our case is the occurrence of a second uterine rupture at a location other than the previous caesarean section scar as well as the previous rupture. The literature reports that almost all reported post-caesarean section ruptures (99%) are located on the old scar [3]. In our case, the only identified risk factor was the surgical history (laparoscopy), which represents a potential iatrogenic risk of occult uterine perforation. Clinical and ultrasound examination did not show signs of adenomyosis or uterine abnormality. Additionally, the patient received genetic counselling by the in-house hospital geneticist consultant. There was no known history of collagen disease in the family. The patient did

Fig. 1. Transabdominal 2D sonography: Hemoperitoneum with free fluid and clots.

Fig. 2. Amniotic sac bulging from the opening of the parietal peritoneum (right) with successive exteriorization of the intact gestational sac and placenta in the abdominal cavity (left).

Fig. 3. Uterine fundal rupture.
not wish to have the recommended laboratory testing for an Ehlers-Danlos syndrome Type IV or Loeys-Dietz syndrome.

The results of the pathology tests for the placenta and the resected myometrium revealed no arguments for a placenta accreta spectrum. The prognosis for a future pregnancy is very challenging. The risk for a third rupture is probably high. We chose to examine the three scars by hydrosonography, which represents an economic and simple examination of the myometrium thickness, but without any qualitative value of the uterine scar. Magnetic resonance imaging is the alternative procedure which allows exploration of the myometrium. We estimated that it would offer no further advantage in this case.

### 4. Conclusion

An increase in the rate of uterine rupture, particularly due to the raised of caesarean sections and labor inductions, has been observed in recent decades [21]. The major causes found in the literature consist of caesarean sections and placental anomalies. As observed in some clinical situations, uterine rupture may remain silent, mainly because of its posterior location which is covered by the omentum or the intestines. In this context, the use of ultrasound plays a key role in the early detection of the rupture, in order to allow for immediate intervention [22]. When possible, the use of an intraoperative two-layer suture is recommended.

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**Table 1**

| Age (years) | Gestity/Parity | Gestational age (GA) | Risk factors | Rupture site |
|-------------|----------------|----------------------|--------------|--------------|
| Aberse et al. [7] | 32 | 2/0 | 21 | Laparoscopy | Fundus |
| Abdalla et al. [8] | 31 | 3/1 | 28 | Curettage | Posterior wall |
| Unzuñ et al. [9] | 32 | 1/0 | 27 | / | Anterior wall |
| Sun et al. [10] | 31 | 3/2 | 17 | / | Fundus |
| Jones [5] | 25 | 0/1 | 21 | Bicornuate uterus | Cornual |
| P.H. Wang et al. [11] | 31 | 1/0 | 21 | Curettage | Cornual |
| P.S. Nagy [12] | 21 | 1/0 | 23 | Placenta percreta | Fundus |
| V. Hlibczuk [13] | 29 | 3/1 | 16 | C-Section | Lower segment |
| G. Ahltoorp [14] | 33 | 1/0 | 16 | Myomectomy | Posterior wall |
| G.R. Damiani et al. [15] | 36 | 2/0 | 23 | Septate uterus | Fundus |
| D. F. Abdulwahab [16] | 36 | 9/8 | 24 | Medical interruption with prostaglandin induction | Anterior wall |
| Torbè et al. [17] | 34 | 4/3 | 21 | Ruptured cornual pregnancy | Fundus |
| H.M. Imseis et al. [18] | 25 | 1/0 | 22 | Myomectomy | Posterior wall |
| Chen [19] | 30 | 1/0 | 26 | Placenta percreta | Fundus |
| M. Nikolaou et al. [20] | 33 | 1/1 | 28 | Adenomyosis | Fundus |
since it can reduce the risk of subsequent ruptures [23]. A risk of recurrent rupture is described up to 22–100% of cases, mainly when it occurs at the fundus [24]. Thus, it is important to consider genetic counselling for patients who have not completed family planning. Based on the World Health Organization (WHO) guidelines, the patient should be advised to wait eighteen months before a new conception. In addition, an elective caesarean section between 36 and 37 weeks of gestation should be offered to the patient according to the guidelines from the American College of Obstetricians and Gynecologists (ACOG). In conclusion, even at an early gestational age, the clinician should consider the possibility of an antepartum uterine rupture in order to reduce maternal and fetal complications.

Contributors
Anne-Claude Fahimi contributed to the literature review and writing of the manuscript. David Salomon contributed to writing of the manuscript. Antonio Zitiello contributed to writing the manuscript. Anis Feki contributed to writing the manuscript. Nordine Ben Ali contributed to project development and writing of the manuscript.

Declarations of Competing Interest
The authors declare that they have no conflict of interest regarding the publication of this case report.

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