Incidence and outcomes of uterine rupture in women with unscarred, preterm or prelabour uteri: data from the international network of obstetric survey systems

Vierin, A.; Vandenberghe, G.; Bloemenkamp, K.; Berlage, S.; Colmorn, L.; Deneux-Tharaux, C.; ... ; INOSS

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RESEARCH ARTICLE

General obstetrics

Incidence and outcomes of uterine rupture in women with unscarred, preterm or prelabour uteri: data from the international network of obstetric survey systems

Griet Vandenberghe[1] | Anne Vierin[1] | Kitty Bloemenkamp[2] | Sylvia Berlage[3] | Lotte Colmorn[4] | Catherine Deneux-Tharaux[5] | Serena Donati[6] | Mika Gissler[7,8,9] | M. Knight[10] | Jens Langhoff-Roos[4] | Pelle G. Lindqvist[11,12] | Barbara Maier[13] | Jos van Roosmalen[14,15] | Joost Zwart[16] | Kristien Roelens[1] | on behalf of INOSS (the International Network of Obstetrics Survey Systems)

[1]Department of Obstetrics & Gynaecology, Ghent University Hospital, Ghent, Belgium

[2]Department of Obstetrics and Gynaecology, Birth Centre Wilhelmina's Children's Hospital, Division Woman and Baby, University Medical Centre Utrecht, Utrecht, the Netherlands

[3]Centre for Quality and Management in Health Care, Medical Association of Lower Saxony, Hannover, Germany

[4]Department of Obstetrics, Rigshospitalet, University Hospital of Copenhagen, Copenhagen, Denmark

[5]Université Paris Cité, Obstetrics, Perinatal and Paediatric Epidemiology Research Team (EPOPé), Centre for Epidemiology and Statistics, Paris, France

[6]National Centre for Disease Prevention and Health Promotion, Istituto Superiore di Sanità (Italian National Institute of Health), Rome, Italy

[7]Department of Knowledge Brokers, THL Finnish Institute for Health and Welfare, Helsinki, Finland

[8]Department of Molecular Medicine and Surgery, Karolinska Institute, Stockholm, Sweden

[9]Academic Primary Health Care Centre, Stockholm, Sweden

[10]National Perinatal Epidemiology Unit, University of Oxford, Oxford, UK

[11]Department of Clinical Science and Education, Karolinska Institute, Stockholm, Sweden

[12]Department of Obstetrics and Gynaecology, Stockholm, Sweden

[13]Department of Gynaecology and Obstetrics, Clinic Ottakring, Vienna Healthcare Group, Vienna, Austria

[14]Athena Institute, VU University Amsterdam, Amsterdam, the Netherlands

[15]Department of Obstetrics, Leiden University Medical Centre, Leiden, the Netherlands

[16]Department of Obstetrics and Gynaecology, Deventer Hospital, Deventer, the Netherlands

Correspondence
G. Vandenberghe, Department of Obstetrics & Gynaecology, Ghent University Hospital, Corneel Heymanslaan 10, Ghent, Belgium.
Email: griet.vandenberghe@uzgent.be

Abstract

Objective: Analysis of atypical cases of uterine rupture, namely, uterine rupture occurring in unscarred, preterm or prelabour uteri.

Design: Descriptive multi-country population-based study.

Setting: Ten high-income countries within the International Network of Obstetric Survey Systems.

Population: Women with unscarred, preterm or prelabour ruptured uteri.

Methods: We merged prospectively collected individual patient data in ten population-based studies of women with complete uterine rupture. In this analysis, we focused on women with uterine rupture of unscarred, preterm or prelabour ruptured uteri.

Main Outcome Measures: Incidence, women's characteristics, presentation and maternal and perinatal outcome.

Results: We identified 357 atypical uterine ruptures in 3 064 923 women giving birth. Estimated incidence was 0.2 per 10 000 women (95% CI 0.2–0.3) in the unscarred

Anne Vierin and Griet Vandenberghe contributed equally to this article.

uteri, 0.5 (95% CI 0.5–0.6) in the preterm uteri, 0.7 (95% CI 0.6–0.8) in the prelabour uteri, and 0.5 (95% CI 0.4–0.5) in the group with no previous caesarean. Atypical uterine rupture resulted in peripartum hysterectomy in 66 women (18.5%, 95% CI 14.3–23.5%), three maternal deaths (0.84%, 95% CI 0.17–2.5%) and perinatal death in 62 infants (19.7%, 95% CI 15.1–25.3%).

Conclusions: Uterine rupture in preterm, prelabour or unscarred uteri are extremely uncommon but were associated with severe maternal and perinatal outcome. We found a mix of risk factors in unscarred uteri, most preterm uterine ruptures occurred in caesarean-scarred uteri and most prelabour uterine ruptures in 'otherwise' scarred uteri. This study may increase awareness among clinicians and raise suspicion of the possibility of uterine rupture under these less expected conditions.

KEYWORDS

caesarean, dehiscence, hysterectomy, pregnancy, prelabour uterine rupture, preterm uterine rupture, scar, unscarred uteri, uterine rupture

1 | INTRODUCTION

Complete uterine rupture is a rare but potentially devastating complication of pregnancy. It represents the complete disruption of the uterine muscle and serosa during pregnancy or childbirth. Immediate intervention is needed to prevent severe asphyxia and perinatal death, while it can necessitate massive transfusion or hysterectomy to save the mother's life.[1–3]

Uterine rupture typically occurs during labour in women at term (≥37 weeks) with a previous caesarean section (CS).[1,4,5] There is a high awareness of clinicians of the risk of uterine rupture when undertaking trial of labour after caesarean (TOLAC), with an estimated incidence of uterine rupture during TOLAC of 35–50/10 000 women based on a firm body of evidence in the literature.[4–9] Clinicians may not expect uterine rupture in an unscarred uterus, prelabour or preterm. However, rupture occurs in 0.3–0.7/10 000 women with unscarred uterus and these ruptures are associated with more severe perinatal and maternal outcomes than with scarred uteri.[4,6,10–12] Uterine rupture may occur before the onset of labour, mostly in scarred uteri, with an estimated incidence of 0.1–0.5/10 000 women based merely on case reports.[6,13–16] Rupture may also occur preterm for various reasons, which has never been a separate focus in the literature.

Atypical ruptures – hereafter referred to as those occurring in unscarred, preterm or prelabour uteri – are very uncommon complications of pregnancy. A multi-country collaboration is the key to facilitate the study of uncommon severe complications by the gathering of a larger number of cases in a shorter timeframe. The International Network of Obstetric Survey Systems (INOSS) merged prospective data of women with complete uterine rupture (disrupted myometrium and serosa) from nine population-based studies.[6–9,17–20] Based on the data of 864 complete uterine ruptures in 2 625 017 deliveries, the INOSS reported an overall incidence of complete uterine rupture of 3.3 (95% CI 3.1–3.5) per 10 000 deliveries, 22 (95% CI 21–24) in women with and 0.6 (95% CI 0.5–0.7) in women without previous CS.[5] The current study focuses on the women with uterine rupture of unscarred uteri, preterm or prelabour uteri only. This unique collection of atypical uterine ruptures over 10 European countries enables the calculation of the estimated incidence of atypical uterine rupture and the description of characteristics and medical factors of these women. This study complements the previously published INOSS study of uterine rupture and together they give a complete overview of all uterine ruptures.

2 | METHODS

2.1 | Research design and data collection

This study is part of a descriptive multi-country population-based study; the research design and data collection have previously been described.[5] Briefly, we merged individual data of women with complete uterine rupture collected prospectively in nine population-based studies: Austria, Belgium, Denmark, Finland, France, Germany, the Netherlands, Sweden and UK. We included extra data from Italy (ItOSS) for this study.[8]

2.2 | Definitions

For the purposes of this study, we defined uterine rupture as the complete disruption of the uterine muscle and serosa, irrespective of the condition of the fetal membranes. Women with asymptomatic dehiscence (intact uterine serosa) noted incidentally during caesarean section (CS) were excluded. Definitions of uterine rupture used by the participating countries have previously been described and are listed in Table S1.[5]

Cases of atypical uterine rupture were subclassified as unscarred, preterm or prelabour. Unscarred was defined as no previous CS or any other uterine surgery prior to the current pregnancy, preterm was defined as gestational age <37^{+0}

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weeks and prelabour was defined as uterine rupture before onset of labour. Labour was defined as having regular uterine contractions and ≥4 cm cervical dilatation (ItOSS) or the diagnosis of labour based on clinical assessment of the reporting physician (other countries). Therefore, women who had induction of labour but not considered to be in labour by their physician, were included in this prelabour group (e.g. there were no regular contractions and there was no change in cervical dilatation).

We created an extra group: the no-previous-caesarean-section group, defined as women without previous CS regardless of their history of other uterine surgery.

Neonatal asphyxia was defined as umbilical artery pH ≤7.00 and/or Apgar score at 5 minutes <7 and/or therapeutic cooling and/or hypoxic ischaemic encephalopathy described as complications.

Perinatal death was defined as intrapartum stillbirths and early neonatal deaths (up to 7 days of life), excluding immature births (gestational age <24 weeks) and termination of pregnancies (due to congenital anomaly or stillbirth prior to uterine rupture).

2.3 | Registered variables

An extensive list of 56 variables was used to make a composite database of all registered variables of atypical cases of uterine rupture. The set of 56 variables and codebook are listed in Table S2. Not every variable was available in all databases. All missing values were coded as not known.

2.4 | Statistical analysis

Data were analysed using statistical software package IBM SPSS statistics for Windows, Version 26.0 (IBM). Most variables were expressed as relative frequencies and statistical analysis was confined to descriptive analyses. Estimated incidences of the different groups were calculated using the estimated total number of women giving birth in the background population during the study period as denominator.

3 | RESULTS

3.1 | Incidence

We identified 357 women who had uterine rupture in an unscarred, preterm or prelabour uterus out of 3 064 923 women who gave birth in 10 European countries. Many of the unscarred ruptures occurred preterm and prelabour. Overlap between the unscarred, preterm and prelabour groups is shown in Figure 1. The overall incidence of atypical uterine rupture was 1.2 (95% CI 1.1–1.3) per 10 000 women who gave birth. The incidence of uterine rupture per 10 000 women who gave birth was 0.2 in the unscarred group (95% CI 0.2–0.3), 0.5 (95% CI 0.5–0.6) in the preterm group, 0.7 (95% CI 0.6–0.8) in the prelabour group and 0.5 (95% CI 0.4–0.5) in the no-previous-CS group. Estimated incidence per country and per group of atypical cases of uterine rupture are shown in Table S3.

3.2 | Women's characteristics

Table 1 provides details of surgical and medical history and obstetric characteristics of women who had an atypical uterine rupture, specified per group of atypical uterine rupture. Three flowcharts presenting women's characteristics per group of atypical uterine rupture further demonstrate the heterogeneity of cases in every group (Figures S1–S3).

There were 75 of 357 women (21%) who had a uterine rupture in an unscarred uterus (no previous CS and no other uterine surgery); 141 women did not have a previous CS but

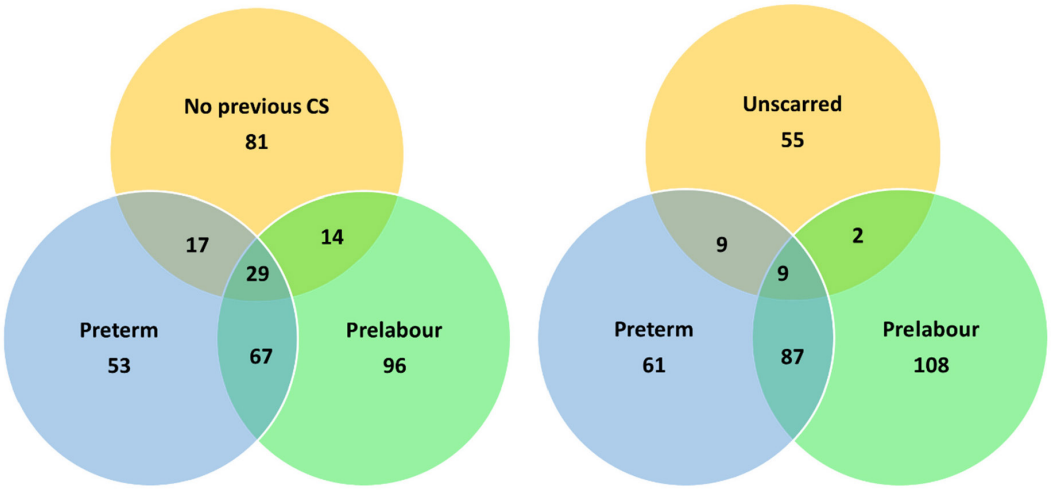


FIGURE 1 Venn diagram showing numbers of uterine rupture of the no-previous-CS or unscarred and/or preterm and/or prelabour uterus, demonstrating overlap in between groups.

TABLE 1 Women's characteristics per group of atypical cases of uterine rupture.

	All atypical cases *n* (%) (*n* = 357)	No-previous-CS *n* (%) (*n* = 141)	Unscarred *n* (%) (*n* = 75)	Preterm *n* (%) (*n* = 166)	Prelabour *n* (%) (*n* = 206)
Surgical history					
Previous uterine surgery	105/337 (31.2)	61/136 (44.9)	0	53/155 (34.2)	69/196 (35.2)
Curettage (D&C)	62	30		22	37
Myomectomy	23	19		19	20
Septal resection	4	4		2	2
Polypectomy	2	2		1	2
Tubectomy	6	4		4	5
Cornual resection	1	0		1	1
B-Lynch	1	1		0	0
Other	5	3		3	3
Previous uterine perforation	7/311 (2.3)	5/124 (4)	0	3/136 (2.2)	6/168 (3.6)
Previous CS	215/356 (60.4)	0	0	119/165 (72.1)	162/205 (79)
1	158			79	118
2	40			29	28
3	11			9	10
4	4			2	4
Number not known	2			0	2
Uterine rupture in history	3/221 (1.4)	0	0	3/114 (2.6)	3/100 (3)
Medical history					
Uterine anomaly[a]	10/214 (4.7)	5/103 (4.9)	1/63 (1.6)	7/109 (6.4)	8/115 (7.0)
Connective tissue disease[b]	1/212 (0.5)	0	0	0	1/115 (0.9)
Adenomyosis/endometriosis	8/212 (3.8)	8/103 (7.8)	1/63 (1.6)	4/109 (3.7)	1/115 (0.9)
Obstetric characteristics					
Labour	146/352 (41.5)	98/141 (69.5)	64/75 (85.3)	65/161 (40.4)	0
Induction of labour	149/357 (41.7)	73/141 (51.8)	42/75 (56)	60/166 (36.1)	76/206 (36.9)
With prostaglandins	130	66	38	56	68
Mechanical	6	0	0	3	3
Oxytocin used (induction/augmentation)	52/270 (19.3)	37/105 (35.2)	26/53 (49.1)	11/133 (8.3)	6/154 (3.9)
TOLAC	104/215 (48.4)	0	0	68/119 (57.1)	56/162 (34.6)
TOP	24/357 (6.7)	7/141 (2.1)	5/75 (6.7)	23/166 (13.8)	5/206 (2.4)
Parity ≥3	47/354 (13.2)	14/138 (10.1)	9/73 (12.3)	26/164 (15.9)	24/205 (11.7)
Multiple pregnancy	9/357 (2.5)	7/141 (4.9)	3/75 (4)	8/166 (4.8)	3/206 (1.5)
Placenta accreta spectrum	12/320 (3.7)	4/132 (3.0)	0	10/154 (6.5)	5/181 (2.7)
Placenta praevia	10/320 (3.1)	1/132 (0.8)	1/70 (1.4)	9/154 (5.8)	9/181 (4.9)
Macrosomia (≥4000 g)	29/320 (9.0)	21/132 (15.9)	17/70 (24.2)	0	11/181
Instrumental vaginal birth	12/357 (3.4)	12/141 (78.7)	7/75 (9.3)	1/166 (0.6)	0

Note: CS, caesarean section; GA, gestational age; IUFD, intrauterine fetal death; TOLAC, trial of labour after caesarean; TOP, termination of pregnancy.

[a]Uterine septum (*n* = 5), uterus arcuatus (*n* = 1), uterus bicornis (*n* = 1), four of unknown type.

[b]Osteogenesis imperfecta type 1.

had a history of other uterine surgery; 44 women had both previous CS and other uterine surgery. The most common types of other uterine surgery were myomectomy, septal resection, tubectomy, resection of cornual pregnancy, polypectomy, curettage and B-lynch suture. There were seven confirmed uterine perforations during dilatation and curettage (D&C) or hysteroscopy. A total of 215 women (60.4%) had at least one previous CS; 55 of them had more than one previous CS; 41 had their previous CS preterm: 8 extreme (24–27 weeks), 10 very (28–31 weeks) and 23 moderate (32–36 weeks) preterm. Three women had recurrent uterine rupture, all three occurred preterm and prelabour.

Most women with unscarred uteri had induced or spontaneous labour in combination with other, mostly obstetric

risk factors (Figure S1). Only six women with unscarred uteri ruptured prelabour: one woman with a ruptured horn of a uterus bicornis at gestational age of 18 weeks, the others with no reported risk factors.

Twelve women with atypical uterine rupture had placenta accreta spectrum (PAS). None of them had unscarred uteri, eight had previous CS and five had other uterine surgery. Ten of these ruptures associated with PAS occurred preterm (range of gestational age 17–35 weeks). They presented with abdominal pain ($n=6$), CTG abnormalities ($n=3$), vaginal bleeding ($n=3$) and altered contractions ($n=2$).

There was one woman with connective tissue disease, osteogenesis imperfecta type 1. Eight women (3.8%) were known to have adenomyosis/endometriosis.

Of 357 women, 166 (46%) had a uterine rupture before 37 weeks. Table 2 shows uterine ruptures stratified per subcategory of preterm birth. Twenty women had a complete uterine rupture at previable gestational age and in 14, uterine rupture occurred during termination of pregnancy (TOP).

3.3 | Presentation

The clinical presentation per type of atypical uterine rupture is demonstrated in Figure 2. The most frequently reported symptoms were abdominal pain (61.6%), abnormal CTG (55.1%) and vaginal bleeding (20.4%).

3.4 | Maternal outcome

Figure 3 shows maternal outcomes per group of atypical uterine rupture. Three maternal deaths occurred, resulting in a case fatality rate of 0.84% (95% CI 0.17–2.5) (Table S4).

In all, 66 of 357 women (18.5%) underwent hysterectomy. Reasons for hysterectomy were massive haemorrhage in 36, severe uterine damage in seven and unclear in 23 women. In 19 women (5.8%) the bladder was damaged and 17 (5.6%) had damage to other organs.

There were 109 women (30.5%) admitted to intensive care units (ICU), with a median stay of 1.7 days (range 1–7), 188 women (55.3%) received some type of blood (packed cells, platelets or plasma) and 114 (32.2%) women were transfused with ≥4 units of packed cells. Fourteen women required intubation and ventilation; one suffered from aspiration pneumonia. Three women had renal dysfunction (one nephrostomy, two renal failures).

Maternal outcomes are presented per type of atypical uterine rupture in Table S4, showing high rates of hysterectomy (30.5%), admission to ICU (44.7%) and massive blood transfusion (51.8%) in the no-previous CS- group and in the unscarred group (26.7%, 42.7% and 50% respectively).

3.5 | Perinatal outcome

This study involved six sets of twins and one triplet, resulting in 365 fetuses. Of these, 274 were born alive: 20 were immature, and 8 were intrauterine fetal deaths, 3 TOPs, 24 peripartum stillbirths, 16 perinatal deaths of unknown timing and 20 fetuses with unknown outcome (Figure S4). There were 62 perinatal deaths (10 antepartum stillbirths, 14 intrapartum stillbirths, 22 neonatal deaths, unknown timing in 16 perinatal deaths). The perinatal mortality rate in atypical uterine rupture was 19.7% (95% CI 15.1–25.3%). Neonatal asphyxia was detected in 35.3% (95% CI 26.9–45.35%). Of the 274 live births, 107 neonates were admitted to neonatal intensive care units (NICU), 35 required intubation/ventilation and seven required therapeutic cooling. Other neonatal complications were intraventricular bleeding ($n=1$), necrotising enterocolitis ($n=2$), hypoxic ischaemic encephalopathy ($n=3$).

Twenty-two neonates died postpartum. Of these, 15 had neonatal asphyxia, two were born extremely preterm (25 and 26 weeks), two were term births with an unclear cause of death, two without neonatal asphyxia had very low birthweights (670 and 633 g) and one had pulmonary hypoplasia caused by anhydramnios.

Figure 4 and Table S5 show perinatal outcome per group of atypical uterine rupture.

4 | DISCUSSION

4.1 | Main findings

Rupture of unscarred, preterm or prelabour uteri is extremely uncommon but is associated with severe maternal and adverse perinatal outcomes. Incidences in this multi-country

TABLE 2 Gestational age per group of atypical cases of uterine rupture.

	All atypical cases *n* (%) (*n* = 357)	No-previous-CS *n* (%) (*n* = 141)	Unscarred *n* (%) (*n* = 75)	Preterm *n* (%) (*n* = 166)	Prelabour *n* (%) (*n* = 206)
Previable (<24 weeks)	20/355 (5.6)	5/141 (3.5)	4/75 (5.3)	20/166 (12)	5/204 (2.4)
Extremely preterm (24–27 weeks)	21/355 (5.9)	6/141 (4.3)	2/75 (2.7)	21/166 (12.7)	16/204 (7.8)
Very preterm (28–31 weeks)	27/255 (7.6)	6/141 (4.3)	1/75 (1.3)	27/166 (16.3)	19/204 (9.2)
Moderate to late preterm (32–36 weeks)	98/255 (27.5)	29/141 (20.6)	11/75 (14.7)	98/166 (59)	56/204 (27.2)
Term (≥37 weeks)	189/255 (52.9)	95/141 (67.4)	57/75 (76)	0	108/204 (52.4)
Not known	2	0	0	0	2

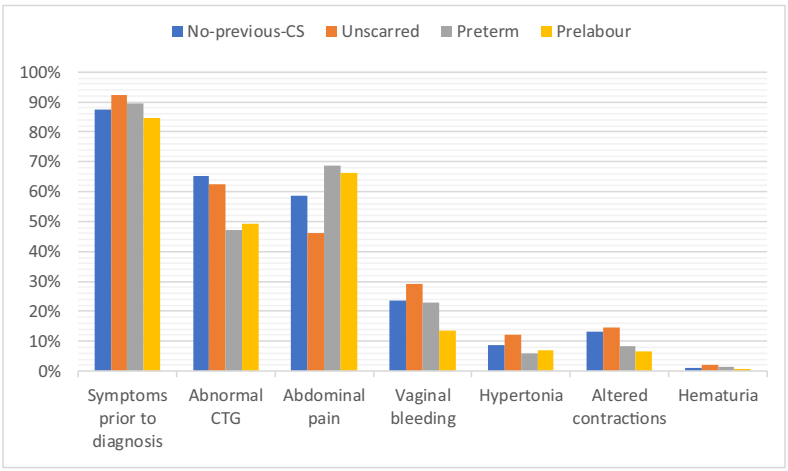
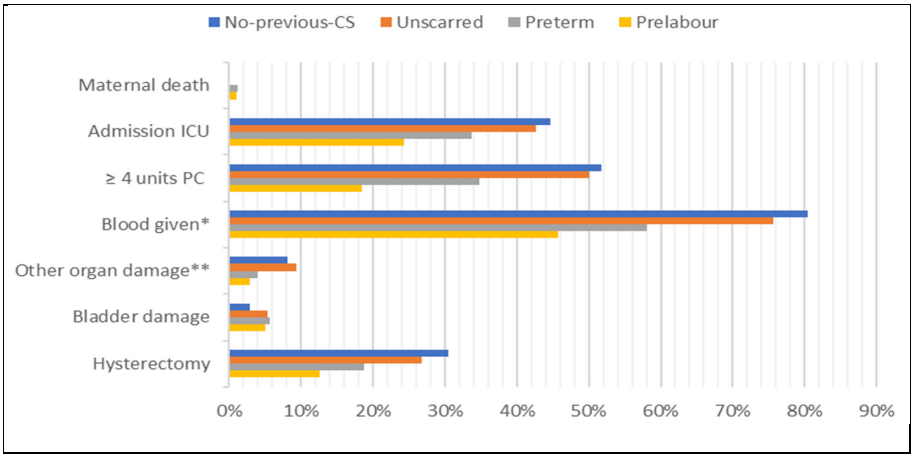


FIGURE 2 Clinical presentation per group of atypical uterine rupture.



PC= packed cells, ICU= intensive care unit
* any type (plasma/packed cells/platelets)
** ureter (n=4), ovaries (n=2), ureter + bowel + ovaries (n=1), broad ligament (n=9) and unknown (n=1)

Other reported maternal morbidity: intubation and ventilation (n=14), aspiration pneumonia (n=1), renal dysfunction (n=3) (one nephrostomy, two renal failures), abscess (n=1), pneumonia (n=1), endometritis (n=1), thrombosis (n=2), multiple organ failure (n=1), diffuse intravascular coagulation (n=3), intracerebral bleeding (n=1), abdominal wound dehiscence (n=2), vesicovaginal fistula (n=1), paralytic ileus (n=1) and visual impairment with spontaneous recovery (n=1).

FIGURE 3 Maternal outcome per group of atypical cases of uterine rupture.

study were 0.2/10 000 in the unscarred, 0.5/10 000 in the preterm, 0.7/10 000 in the prelabour and 0.5/10 000 in the no-previous-CS group. Atypical uterine rupture commonly resulted in peripartum hysterectomy, ICU admission and massive transfusion in 31%, 45% and 52%, respectively. Although maternal death occurred in only 0.8%, neonatal asphyxia (35%) and perinatal mortality (20%) were common.

4.2 | Strengths and limitations

The main strength of this study is its multi-country population-based design. Thanks to the INOSS collaboration, we assembled a unique collection of 357 women with complete uterine rupture in unscarred, preterm or prelabour uteri in over 10 European countries.[5] This large number of atypical uterine ruptures improves the understanding of obstetricians: in which clinical situations should they be more vigilant for this uncommon complication?

The main limitations are the differences in collected variables between countries, especially variables concerning medical history and perinatal outcome. This demonstrates the importance of the set of core outcome measures developed by INOSS for future studies.[21] Information on type of uterine incision in previous CS and gestational age of the previous CS were largely missing; this could have provided more insight in uterine rupture risk when previous CS was very preterm. Some information, however, may be unknown even by reporting physicians, such as previous TOP and uterine perforation during D&C.

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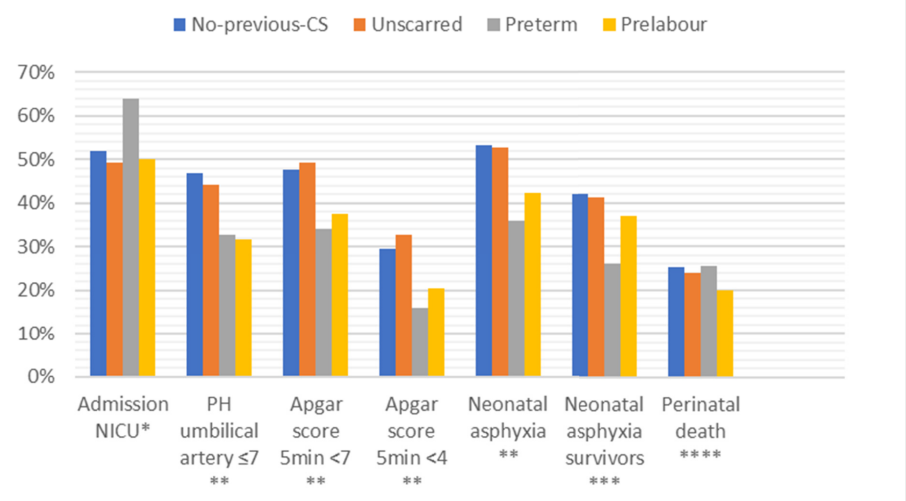

NICU=neonatal intensive care unit
* without 55 not applicable cases (20 immature + 11 TOP + 10 antepartum deaths + 14 intrapartum deaths)
** without 41 not applicable cases (20 immature + 11 TOP + 10 antepartum deaths)
*** with extraction of perinatal deaths
**** without immature + TOP + unknown timing of perinatal death

FIGURE 4 Perinatal outcome per group of atypical cases of uterine rupture.

Lack of information existed on the distribution of surgical, medical and obstetric characteristics in the general population of pregnant women. As we could not test for specific features of women with atypical uterine rupture, we were confined to descriptive analysis. Estimated incidence could only be calculated using the total number of women giving birth in the study period as the denominator.

An important limitation is lack of an objective definition for labour in most population-based studies. As a consequence, incidence of uterine rupture in the prelabour uterus may be overestimated. Moreover, 76 women in this prelabour group underwent induction of labour, most of them (68) with prostaglandins. If we exclude these, the incidence comes down from 0.7 to 0.4/10 000 women giving birth.

Finally, we cannot provide data on timing of diagnosis and decision-to-delivery interval.

4.3 | Interpretation

The term unscarred uterus is discordantly used in the literature: no previous CS, no previous uterine surgery involving myometrial incision or no uterine surgery at all. Therefore, we distinguished between unscarred (without any uterine surgery) and no-previous-CS uteri (without previous CS regardless of other uterine surgery) in this study.

Our incidences are in line with the literature. In Norway, 65 uterine ruptures (0.45/10 000) occurred in intact uteri among 1 441 712 women giving birth.[4] Primary uterine rupture (defined as no history of uterine surgery involving myometrial incision) occurred in Utah, USA, in 0.45/10 000 women giving birth and in Denmark in 0.3/10 000 women without previous CS.[10,11]

Almost half of the women in the no-previous-CS group (44.9%) had other previous uterine surgery. A history of myomectomy was reported in 23 women with uterine rupture, most occurring preterm and prelabour. A literature review in 2007 reported previous myomectomy in 11 of 36 primigravid women with uterine rupture.[12] Association of uterine rupture with septal resection, operative hysteroscopy, utero-tubal surgery, D&C and B-lynch suture is rarely documented. In 2006, 18 women were reported with uterine rupture after operative hysteroscopy.[22] Seven uterine perforations occurred during D&C or hysteroscopy in our study; however, uterine perforations may go unnoticed or stay unreported by physicians. We plead for increased carefulness by gynaecologists to detect, document and report uterine perforations at all times.

But then, why do unscarred uteri rupture? This large group demonstrated a mix of risk factors previously associated with uterine rupture: grand multiparity, multiple gestation, macrosomia, uterotonics, instrumental birth, manual removal of placenta, PAS, congenital uterine anomaly, adenomyosis and connective tissue disease.[10,12,23–26] Other risk factors not present in our study were external trauma, traffic accidents, previous invasive molar pregnancy, uterine diverticula, chronic corticosteroid use or cocaine abuse. Instrumental vaginal birth could be either management or cause of uterine rupture from trauma, although this was extremely rare. Especially in cases with fetal distress it may be difficult to distinguish whether rupture occurred before or during birth. The woman with connective tissue disease had osteogenesis imperfecta type 1, with a less known risk of uterine rupture than Ehlers–Danlos. We found four more case reports of uterine rupture in women with osteogenesis imperfecta type 1.[27–30]

Why do uteri rupture preterm? The majority had a previous CS and a vast number occurred during labour. Twenty-four occurred during TOP due to antepartum stillbirth or congenital anomaly. Most preterm ruptures occurred in a combination of medical and obstetric risk factors. Only 18 preterm uteri were unscarred. We found no studies addressing preterm uterine rupture in literature.

Data on prelabour uterine rupture consist mainly of case reports. In Norway, 0.03/10 000 occurred in unscarred uteri (8 in 2 334 712) and 1.8/10000 in scarred uteri (22 in 121 085) giving an incidence of prelabour uterine rupture of 0.1/10 000 women giving birth.[13] A study of the Netherlands reported an incidence of 0.5/10 000 women giving birth.[6]

Why do prelabour uteri rupture? Most women had a previous CS or other uterine surgery, in accordance with the literature.[6,13–16] All three recurrent uterine ruptures, and the majority of uterine ruptures in women with previous myomectomy (20/23), occurred prelabour. In a review by Uccella et al., 16 of 24 primigravid women with prelabour uterine rupture had previous uterine surgery (mostly myomectomies).[16] A systematic review reported in 2016 that uterine rupture after myomectomy occurred even when the uterine cavity was not opened, mainly before 36 weeks and before labour ($n = 5/330$, 1.5%); only 2/426 (0.47%) occurred during a trial of labour after myomectomy.[31]

Consequences of atypical uterine rupture were severe, with high rates of hysterectomy and poor perinatal outcomes, coherent with previous studies reporting more severe outcome associated with uterine rupture in unscarred uteri.[6,10] This may be explained by delayed diagnosis and management due to the unexpectedness. Also, higher risks of massive haemorrhage could be involved in the more vascular unscarred myometrium compared with uterine scar tissue.[7]

5 | CONCLUSION

Uterine rupture in preterm, prelabour or unscarred uteri is extremely uncommon but is associated with severe maternal and perinatal outcomes. We found a mix of risk factors in unscarred uteri, and most preterm uterine ruptures occurred in CS-scarred uteri and most prelabour uterine ruptures in 'otherwise' scarred uteri. Clinicians should carefully document any other uterine surgery and women should be counselled of the potential rare but catastrophic risks of uterine rupture, even at preterm gestational age. This study may increase the awareness of clinicians and raise suspicion of the possibility of uterine rupture under these less expected conditions.

AUTHOR CONTRIBUTIONS

GV designed the study and coordinated data collection. AV coordinated data processing and analysis. All authors contributed in the interpretation of the data. AV and GV drafted the paper. All authors critically revised the article and approved the final version.

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ETHICS APPROVAL

None.

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CONFLICT OF INTEREST STATEMENT

None declared.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding authors. The data are not publicly available due to privacy or ethical restrictions.

ETHICAL APPROVAL

Because of the character of this study, ethical approval was not required. The use of each study from which data were used, was approved by their national or local ethics committee.

ORCID

Griet Vandenberghe https://orcid.org/0000-0002-7420-5020
Catherine Deneux-Tharaux https://orcid.org/0000-0002-6561-3321
Serena Donati https://orcid.org/0000-0002-4607-2072
Marian Knight https://orcid.org/0000-0002-1984-4575
Pelle G. Lindqvist https://orcid.org/0000-0002-1652-8235
Jos van Roosmalen https://orcid.org/0000-0002-6746-3259

REFERENCES

1. Hofmeyr GJ, Say L, Gülmezoglu AM. WHO systematic review of maternal mortality and morbidity: the prevalence of uterine rupture. BJOG: An International Journal of Obstetrics and Gynaecology. 2005;112(9):1221–8.
2. Al-Zirqi I, Daltveit AK, Vangen S. Infant outcome after complete uterine rupture. American Journal of Obstetrics and Gynecology. 2018;219(1):109.e1–8.
3. Al-Zirqi I, Daltveit AK, Vangen S. Maternal outcome after complete uterine rupture. Acta Obstetricia et Gynecologica Scandinavica. 2019;98(8):1024–31.
4. Al-Zirqi I, Stray-Pedersen B, Forsén L, Daltveit AK, Vangen S. Uterine rupture: trends over 40 years. BJOG. 2016;123(5):780–7.
5. Vandenberghe G, Bloemenkamp K, Berlage S, Colmorn L, Deneux-Tharaux C, Gissler M, et al. The international network of obstetric survey systems study of uterine rupture: a descriptive multi-country population-based study. BJOG. 2019;126(3):370–81.
6. Zwart JJ, Richters JM, Ory F, de Vries JI, Bloemenkamp KW, van Roosmalen J. Uterine rupture in The Netherlands: a nationwide population-based cohort study. BJOG. 2009;116(8):1069–78. discussion 1078–80.

7. Vandenberghe G, De Blaere M, Van Leeuw V, Roelens K, Englert Y, Hanssens M, et al. Nationwide population-based cohort study of uterine rupture in Belgium: results from the Belgian obstetric surveillance system. BMJ Open. 2016;6(5):e010415.
8. Donati S, Fano V, Maraschini A. Uterine rupture: results from a prospective population-based study in Italy. European Journal of Obstetrics, Gynecology, and Reproductive Biology. 2021;264:70–5.
9. Fitzpatrick KE, Kurinczuk JJ, Alfirevic Z, Spark P, Brocklehurst P, Knight M. Uterine rupture by intended mode of delivery in the UK: a national case–control study. PLoS Med. 2012;9(3):e1001184.
10. Gibbins KJ, Weber T, Holmgren CM, Porter TF, Varner MW, Manuck TA. Maternal and fetal morbidity associated with uterine rupture of the unscarred uterus. American Journal of Obstetrics and Gynecology. 2015;213(3):382.e1–6.
11. Thisted DL, Mortensen LH, Krebs L. Uterine rupture without previous caesarean delivery: a population-based cohort study. European Journal of Obstetrics, Gynecology, and Reproductive Biology. 2015;195:151–5.
12. Walsh CA, Baxi LV. Rupture of the primigravid uterus: a review of the literature. Obstet Gynecol Surv. 2007;62(5):327–34. quiz 53–4.
13. Al-Zirqi I, Vangen S. Prelabour uterine rupture: characteristics and outcomes. BJOG. 2020;127(13):1637–44.
14. Abbas AM, Michael A, Ali SS, Makhlouf AA, Ali MN, Khalifa MA. Spontaneous prelabour recurrent uterine rupture after laparoscopic myomectomy. J Obstet Gynaecol. 2018;38(7):1033–104.
15. Odejinmi F, Annan J, Wong I. Pre-labour spontaneous uterine rupture at 31 weeks' gestation following laparoscopic excision of a broad ligament paramesonephric cyst and literature review. J Obstet Gynaecol. 2011;31(5):444–5.
16. Uccella S, Cromi A, Bogani G, Zaffaroni E, Ghezzi F. Spontaneous prelabor uterine rupture in a primigravida: a case report and review of the literature. American Journal of Obstetrics and Gynecology. 2011;205(5):e6–8.
17. Knight M. The international network of obstetric survey systems (INOSS): benefits of multi-country studies of severe and uncommon maternal morbidities. Acta Obstetricia et Gynecologica Scandinavica. 2014;93(2):127–31.
18. Berlage S, Grüßner S, Lack N, Franz HB. GerOSS (German obstetric surveillance system). A project to improve the treatment of obstetric rare diseases and complications using a web based documentation and information platform. Methods Inf Med. 2015;54(5):406–11.
19. Colmorn LB, Petersen KB, Jakobsson M, Lindqvist PG, Klungsoyr K, Källen K, et al. The Nordic obstetric surveillance study: a study of complete uterine rupture, abnormally invasive placenta, peripartum hysterectomy, and severe blood loss at delivery. Acta Obstetricia et Gynecologica Scandinavica. 2015;94(7):734–44.
20. Deneux-Tharaux C, Bouvier-Colle M-H. 585: severe acute maternal morbidity in France: the epimoms population-based study. American Journal of Obstetrics and Gynecology. 2017;216(1):S345–6.
21. Schaap T, Bloemenkamp K, Deneux-Tharaux C, Knight M, Langhoff-Roos J, Sullivan E, et al. Defining definitions: a Delphi study to develop a core outcome set for conditions of severe maternal morbidity. BJOG. 2019;126(3):394–401.
22. Sentilhes L, Sergent F, Berthier A, Catala L, Descamps P, Marpeau L. Uterine rupture following operative hysteroscopy. Gynecol Obstet Fertil. 2006;34(11):1064–70.
23. Ofir K, Sheiner E, Levy A, Katz M, Mazor M. Uterine rupture: differences between a scarred and an unscarred uterus. American Journal of Obstetrics and Gynecology. 2004;191(2):425–9.
24. Mazzone ME, Woolever J. Uterine rupture in a patient with an unscarred uterus: a case study. WMJ. 2006;105(2):64–6.
25. Faria J, Henriques C, Silva Mdo C, Mira R. Rupture of an unscarred uterus diagnosed in the puerperium: a rare occurrence. BMJ Case Rep. 2012;21:bcr2012006372.
26. Turner MJ. Uterine rupture. Best Pract Res Clin Obstet Gynaecol. 2002;16(1):69–79.
27. Redman MG, Wagner BE, Balasubramanian M. Osteogenesis imperfecta type I: the role of deep phenotyping in a patient with a ruptured uterus. Eur J Med Genet. 2020;63(12):104095.
28. Di Lieto A, Pollio F, De Falco M, Iannotti F, Mascolo M, Somma P, et al. Collagen content and growth factor immunoexpression in uterine lower segment of type IA osteogenesis imperfecta: relationship with recurrent uterine rupture in pregnancy. American Journal of Obstetrics and Gynecology. 2003;189(2):594–600.
29. Krishnamoorthy U, Vausse S, Donnai P. Management of pregnancy complicated by maternal osteogenesis imperfecta. Report of a case with uterine rupture. J Obstet Gynaecol. 2002;22(3):316.
30. Christodoulou S, Freemont AJ, McVey R, Vause S. Prospective comparative case study of uterine collagen in a woman with osteogenesis imperfecta type 1 who had previously ruptured her uterus. J Obstet Gynaecol. 2007;27(7):738–9.
31. Gambacorti-Passerini Z, Gimovsky AC, Locatelli A, Berghella V. Trial of labor after myomectomy and uterine rupture: a systematic review. Acta Obstetricia et Gynecologica Scandinavica. 2016;95(7):724–34.

SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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