A CASE REPORT OF DELAYED DIAGNOSIS OF UTERINE RUPTURE FOLLOWING VAGINAL DELIVERY

Vanessa Falé Rosado[*,1], **Sara Rocha**[*], **Catarina Vasconcelos**[*], **Maria Luisa Martins**[*] **and Maria José Alves**[*]
[*]Maternidade Dr. Alfredo da Costa, Rua do Viriato, Lisbon, Portugal.

ABSTRACT Introduction: A uterine rupture is still a rare event, but its incidence appears to be increasing, even in the unscarred uterus. In our case, the uterine rupture presented itself in an unscarred uterus and after a vaginal delivery. **Case report:** A 36 years old women with three previous normal deliveries, comes to our hospital for assistance at 32 weeks with a poor pregnancy surveillance. After diagnosing Gestational Diabetes, she is admitted for therapeutic adjustment. She is discharged after achieving metabolic control but comes back a few days later with a stillbirth, born by vaginal delivery. Six days later she presents with: fever and pain; anaemia leukocytosis and a heterogeneous image on ultrasound. However, was decided to start intravenous antibiotics before choosing for surgery. Her condition worsens, and an exploratory laparotomy is done: a posterior uterine wall rupture that required a hysterectomy. **Conclusion:** Risk factors for uterine rupture were present (maternal age over 35, higher parity, fetal macrosomia) but the absence of any symptom, the regular examination after delivery, and mostly, an unscarred uterus, resulted in a delay in the diagnosis of more than one week, leading to catastrophic consequences: hysterectomy. This case reminds us that uterine rupture happens not only in case of previous uterine surgery, and these cases seem to be increasing because of the increase in other risk factors: advanced maternal age and diabetes with resulting fetal macrosomia.

KEYWORDS uterine rupture, hysterectomy, postpartum period

Introduction

Uterine rupture represents a breach in the integrity of the uterine muscle, usually during labour although it can also happen before. It has been considered an entity with dramatic consequences both for the mother and the fetus, leading to severe haemorrhage on the mother and hypoxia in the fetus. Because it can have no symptoms, the diagnose may be delayed aggravating the prognosis. This obstetric emergency, considered rare, seems to be increasing due to the rising rate of uterine surgeries like cesareans and myomectomies (due to the delaying in maternity). However, in our case, no previous uterine surgery was reported. This aspect combined with the lack of any symptom, resulted in a delay of more than one week in the diagnosis, which in turn led to disastrous consequences - hysterectomy. Better methods for predicting this disaster complication are needed, especially considering its increasing incidence also in the unscarred uterus, possibly due to the growing of other risk factors like advanced maternal age and gestational diabetes with fetal macrosomia.

DOI: 10.5455/IJMRCR.uterine-rupture-delayed-diagnosis
First Received: July 07, 2017
Accepted: July 22, 2017
Manuscript Associate Editor: George Baytchev (BG)
Editor-in Chief: Cvetanka Hristova (BG)
Reviewers: Ivan Inkov (BG)
[1]Maternidade Dr. Alfredo da Costa, Rua do Viriato, Lisbon, Portugal., vanessasfrosado@hotmail.com

Case report

A thirty-six years old woman, melanodermic, natural from Africa, came to Lisbon, Portugal at 32 weeks of pregnancy to have the remaining pregnancy surveillance and delivery in Maternidade Dr Alfredo da Costa. Her personal and family background were irrelevant. As for previous deliveries, she mentioned three eutopic deliveries: in 2002 a healthy baby with 3800grs, in 2004 she referred a late premature delivery of a healthy baby with 3400grs and finally in 2014, a pregnancy complicated with gestational diabetes that resulted in a newborn

Table 1 Analysis of patients' clinical evolution on antibiotic therapy

	Day 0	Day 1	Day 2	Day 3	Day 5
Hb (gr/dL)	9,8	8,3	8,8	7,8	8,1
WC (cells/μL)	26 180	17 500	16 500	13 000	17 200
N (%)	91	89	88	84	86
CRP (mg/L)	397	316	315	239	260
US image size	105x95x70	116x99x59			

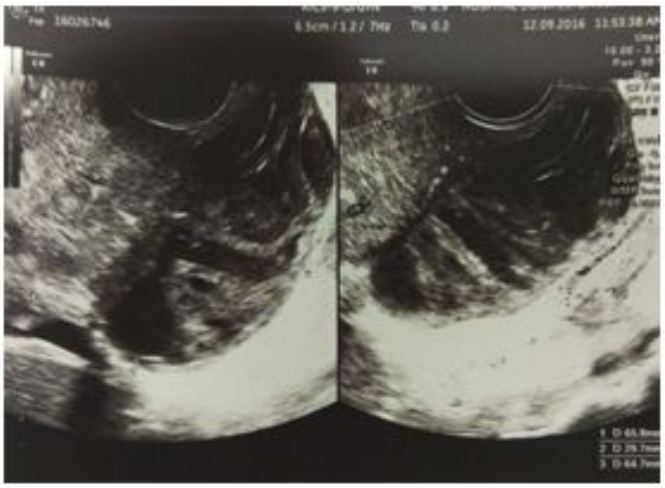

Figure 1: Ultrasound image at emergency department, eight days after delivery (U – uterus; HI – heterogeneous image)

with 3200grs. A reclassification test of gestational diabetes was not done. All pregnancies were with the same partner. In her first appointment in Lisbon at $32^{+}5$ weeks, it was decided to repeat a glucose tolerance test (75grs glucose with glycaemia measurement at 0, 1 and 2 hours). The results were positive: 191, 239 and 339 mg/dL. An ultrasound was performed: the fetus was in the 95^{th} percentile (Hadlock et al) and the amniotic fluid level was normal. She was admitted to the hospital for maternal-fetal surveillance, and glycemic control was achieved with diet, insulin and metformin. She was released at $37^{+}1$ with good metabolic control; a new appointment and elective cesarean for 39 weeks were scheduled.

At 38 weeks she came to the emergency department because of the absence of fetal movements for the last 12 hours: she was diagnosed with stillbirth. She went into spontaneous labour, and 10 hours later, after a breech delivery, she had a stillborn with 4525 grs. The family refused an autopsy.

After 48hrs of inpatient observation, she was discharged from the hospital.

She came back to the emergency department six days later with complaints of fever, pelvic pain and foul smelling vaginal discharge, which was confirmed by observation. On ultrasound, an image with heterogeneous echogenicity was found over the uterine fundus, measuring 105x95x70 mm. Blood analysis revealed low levels of haemoglobin (9,8gr/dL), leukocytosis (26 180/μL), neutrophilia (91%) and elevated C-reactive protein (397mg/L). She was initiated on endovenous antibiotic therapy with gentamycin, ampicillin and clindamycin. Analytic evolution is present in table 1.

Because of clinical deterioration, exploratory laparotomy was

Table 2 Timeline of the patient.

Dates	**Relevant Past Medical History and Interventions**		
2002	Vaginal delivery in Africa, newborn with 3800 grs, without any interferences during pregnancy, intrapartum or postpartum		
2004	Vaginal delivery in Africa, newborn with 3400 grs, without any interferences during pregnancy, intrapartum or postpartum		
2014	Vaginal delivery in Africa, newborn with 3200 grs, diagnosis of Gestational Diabetes without posterior reclassification test		
Dates (in 2016)	**Summaries from Initial and Follow-up Visits**	**Diagnostic Testing (including dates)**	**Interventions**
22 July	The first appointment in our hospital; none of the previous exams done in Africa, were available for consultation	Ultrasound was done in Portugal on 18 July revealing a healthy baby with 32+5 weeks	Analysis with 75grs glucose tolerance test was requested
12 August	35+5 weeks Asymptomatic	-Glucose tolerance test positive: 191/238/339 -Ultrasound that day: fetus on 95th percentile	Admission to Maternal-fetal Infirmary for surveillance and therapeutic adjustment with both insulin and metformin
22 August	37+1 weeks Asymptomatic	Adequate metabolic control and daily cardiotocography showing fetal well being	Cesarean scheduled to 6 of September
30 August	38+2 weeks No fetal movements	Stillbirth on ultrasound	Breech delivery 10 hours later: Dead fetus with 4525 grs
1 September	Asymptomatic Clinical observation normal: normal lochia, normal uterine involution and painless examination		48hrs after delivery, she is discharged from Hospital with metabolic control
7 September	8th day after delivery Fever, pelvic pain and foul smelling vaginal discharge, which was confirmed by observation	-Ultrasound: image with heterogeneous echogenicity was found over the uterine fundus, with 105x95x70 mm -Anemia(9,8gr/dL), leukocytosis (26 180/μL), neutrophilia (91%) and elevated C-reactive protein (397mg/L)	Admitted to hospital and initiated ampicillin, gentamicin and clindamycin IV
8-10 September	Improvement of symptoms and clinical observation: pyrexia, no pain at fundal uterine palpation	-Blood test improvements, -Ultrasound with stable image (without increases in size)	Maintain therapeutic
12 September	13th day after delivery 5th day with antibiotic Clinical deterioration: fever, pain	Deterioration of both blood test and ultrasound image	Exploratory laparotomy: vast uterine rupture of posterior wall and a hysterectomy was decided
10 October			Discharged from hospital after therapeutic with meropenem, during three weeks because of multiresistant Klebsiella pneumoniae isolated in the purulent exudate collected from the abdominal cavity during, surgery

Table 3 Medical History of eight cases of rupture (CS – cesarian; Leuco – leucodermic; melano – melanodermic; leucomelano - leucomelanodermic)

			Obstetric and Gynecological History						
#	Race	Age	Dilatation & Curettage	Nulliparous	$\geq$ 3 Deliveries	1 CS	$\geq$ 2 CS	Time since last cesarian (in months)	Other uterine surgery
1	Leuco	31	No	No	No	Yes	No	21	No
2	Melano	31	Yes (2)	No	No	Yes	No	48	No
3	Melano	29	No	No	No	Yes	No	3	No
4	Leuco	44	Yes (1)	Yes	No	No	No	-	No
5	Melano	26	No	No	No	No		15	No
6	Leucomelano	29	Yes (3)	No	No	Yes	No	84	laparotomic cerclage
7	Melano	36	No	No	Yes		No	-	No
8	Leuco	37	No	No	No	Yes	No	72	No

decided. On surgical exploration a large rupture was found on the posterior uterine wall; there was purulent exudate all over the abdominal cavity. A total hysterectomy was performed with unilateral adnexectomy due to technical difficulties.

After surgery, this patient stayed in the hospital for almost one month due to the isolation of a multiresistant Klebsiella Pneumoniae, which responded to Meropenem.

The mean age of this small sample was 33 years old (6 STD; min 26, max 44). Half were melanodermic and six had a previous cesarean. No case of prior myomectomy. Most went into spontaneous labour and most had a cesarean.

As for the rupture itself, some situations were identified as dehiscence and not as a complete disruption of the uterine wall. This means that the serosa was still intact. The breach location was mostly on the previous historiography, therefore on the anterior uterine wall.

Finally, the timing of the diagnosis was mainly intrapartum; only one situation was diagnosed before, during the early third trimester, with inferior segment measurement on ultrasound; and the already mentioned rupture, that was diagnosed seven days after delivery.

Using the International Classification of Diseases 9^{th} Edition (ICD 66501 and 6651), we identified eight cases of uterine rupture in our hospital from January 2010 to September 2016 out of a total of 29 797 (average rate of cesarean of 28,5%).

Discussion

The clinical significance of uterine rupture is growing, as its prevalence appears to be rising. However, it represents many challenges for modern obstetrics: its risk factors identification, its timely diagnosis and its prevention.

Many authors consider two types of uterine rupture: dehiscence and total rupture. Rupture would represent a disruption of both uterine muscle and visceral peritoneum while in dehiscence the peritoneum is intact.[1] Some authors prefer to consider dehiscence a partial rupture as opposed to total rupture.[2]

The incidence is somewhat difficult to define due to the scarcity of good quality studies since most of them are case reports. For this reason, uterine rupture rate varies from 1/10,000 to 1/16,840-19,765, in the unscarred uterus. In the event of a previous cesarean, then the rate goes higher as expected: 1/1235-4366 and as high as one /100. [3-8]

Intrapartum diagnosis poses many obstacles as Holmgren et al. showed in a study of 36 cases of rupture.[9] This study shows which clinical sign led to the decision to perform a cesarean in a woman attempting trial of labor after cesarean (TOLAC): 30,5% severe variable decelerations, 19,4% prolonged fetal bradycardia, 22% maternal symptoms like pain and hypotension with no fetal heart rate concerning changes. The maternal pain was present in 25% of patients. This means that symptomatology is very varied and no symptom appears to be more frequent. This difficulty in intrapartum diagnosis increases time to delivery with possibly serious consequences for the mother and neonate According to this group, every additional minute to delivery enhances the risk of a neonatal adverse outcome by 8,8%. As for risk assessment, many clinical factors have been pointed out: maternal age equal or higher than 35 years, parity equal or higher than three, non-Western maternal origin, use of oxytocin, prostaglandins or transcervical balloon, scarred uterus (multiple previous cesarean section and type of previous hysterotomy closure), fetal macrosomia (birth weight $\geq$ 4 kg).[10-12] The one that reunites most agreement is the classical cesarean. In a study from 2012 by Gyamfi-Bannerman et al., three groups were compared: prior myomectomy, prior classical cesarean and previous lower segment transverse cesarean.[13] The main conclusions were that classical cesarean increased the risk of uterine rupture (adjusted OR 3,23), while previous myomectomy had no risk. Despite these results, the group of prior myomectomy had some important differences from the groups of classical cesarean and lower segment transverse cesarean: lower rate of induction (1,1% vs. 2,6% for classical and 17,8% lower segment), lower rate of vaginal delivery (0% vs. 5,9% for classical and 44,2% for lower segment).

What is emerging, as a possibility to predict this disastrous

Table 4 Resume of type of delivery, type of rupture and neonatal outcomes of eight cases (time in labour in hours; CS – cesarian; N.B.: newborn; AS – Apgar Score)

	Labor		Rupture Type			Delivery				
#	Spontaneous\ induction/ cesarian	Time in labor	Timing of diagnosis	Type	Site of uterine disruption	Other complications	Type	Weeks	NB weight	AS
1	spontaneous	4	intrapartum	dehiscence	anterior wall	No neonatal intercorrence	CS	38	2755	9//10
2	spontaneous	1	intrapartum	rupture	broad ligament & lateral wall	No neonatal intercorrence	CS	39	2960	9//9
3	spontaneous	2	intrapartum	dehiscence	anterior wall	No neonatal intercorrence	CS	39	3870	9//10
4	induction	6	intrapartum	rupture	posterior wall	No neonatal intercorrence	CS	38	3000	9//10
5	elective cesarian		intrapartum	dehiscence	anterior wall	No neonatal intercorrence	CS	39	3450	9//10
6	spontaneous	3	intrapartum	rupture	anterior wall	No neonatal intercorrence	CS	35	2230	9//10
7	spontaneous	2	postpartum	rupture	posterior wall	Hysterectomy; stillbirth	Breech	38	4525	0//0
8	spontaneous	5	prepartum	dehiscence	anterior wall	No neonatal intercorrence	CS	31	1580	9//9

complication, is lower uterine segment (LUS) measurement in women attempting TOLAC.

According to Bujold et al., the measurement should be done between 35 and $38^{+}6$ weeks, using both the transvaginal and the transabdominal probes, to reduce interobserver variability.[14] Three measurements should be done and the smaller considered. However, many debates surround this theme as for what should be measured and what the right cut-off value to consider. For some authors, full segment thickness is more predictive of uterine scar rupture. This means measuring both the myometrium and the bladder wall. However in a recent meta-analysis by Kok et al[15] measuring only the myometrium is as useful in predicting rupture. Another issue to debate is cut-off value: it has varied from 3,5 to 2 mm, and so far no precise value can be recommended.[16]

In this case report the rupture occurred in an intact uterus, which makes it even harder to predict and diagnose and therefore with worst consequences as shown in a retrospective study by Gibbins et al.[17] After exclusion of prednisone chronic use, connective tissue disease and multiple pregnancies, 146 cases of uterine rupture were selected; 20 of them in intact uterus. After comparing these two groups (intact vs. scarred uterus), the major difference is the delay in the uterine rupture diagnosis: in women with a previous uterine scar, the delivery was more often a cesarean (91% vs. 58%), and for this reason the diagnosis was made intrapartum during the cesarean; as for intact uterus almost half women had a vaginal delivery and the diagnosis was delayed resulting in higher blood loss (2000 vs. 800mL, p <0,001), hysterectomy rates (35 vs. 2,4%, p < 0,001) and maternal morbidity (65 vs. 20%, p < 0,001).

As mentioned above, every obstetrician should be more and more aware of this condition because recent analysis shows its increasing incidence like this Norwegian study from 2015.[12] Deliveries from 21 hospitals from 1967 to 2008 were selected and divided into four groups that correspond to four decades: 67-77, 78-88, 89-99, 00-08. The incidence increased abruptly in the last decade: from 0.9/10 000 in the second decade (1978–1988) to 6.1/10 000 in the fourth decade (2000–2008). This increase was mostly because of the scarred uterus group: 14.2/10 000 in the second decade to 66.8/10 000 in the fourth decade. Indeed, scarred uterus and labour augmentation with oxytocin were the main contributors to this increase in uterine rupture. In the intact uterus, after adjusting for prostaglandins and oxytocin use, the OR remains almost the same.

Finally, after a uterine rupture should we be tremendously scared of the next pregnancy? In a small study by Fox et al. with 60 pregnancies (20 after uterine rupture and 40 after uterine dehiscence), pregnancy outcomes were very good with no cases of repeated rupture or hysterectomy. [18] The only dehiscence repeated itself: 5% in the rupture group and 7,5% in the dehiscence group. All pregnancies were ended before 40 weeks: in the rupture group 75% ended between 36 and 37 weeks via cesarean and in the dehiscence group 93% between 36 and 39 weeks via cesarean.

Conclusion

Uterine rupture is a serious complication that can be more easily suspected in the case of the previous cesarean; however, it can also happen in the intact uterus and, even though it is a rare event, factors like labour augmentation and prostaglandins use for labour induction can eventually lead to an increase in its prevalence.

In this particular case, some risk factors can be identified as fetal macrosomia, higher parity and maternal age over 35. Still, she was discharged from the hospital 48hrs after delivery with no complaints and after a normal examination. Only one week later she comes back with pain and fever. This delay in diagnosis of more than one week after delivery led to very serious morbidity like mentioned above by Gibbins et al.

For all that it was mentioned, uterine rupture should be a diagnostic hypothesis to consider, especially if risk factors like older maternal age and previous uterine scar are present. An earlier clinical suspicion means a better obstetric outcome.

Authors' Statements

Competing Interests

The authors declare no conflict of interest.

References

1. Landon MB, Hauth JC, Leveno KJ, et al. Maternal and perinatal outcomes associated with a trial of labour after prior cesarean delivery. N Engl J Med 2004;351: 2581-9.

2. Nahun GG, Pham KQ. Uterine rupture in Pregnancy. Medscape Reference, 25 Mar, 2016 [www.reference.medscape.com/article/275854-overview]. Accessed 2 March 2017.

3. Gardeil F, Daly S, Turner MJ. Uterine rupture in pregnancy reviewed. Eur J Obstet Gynecol Reprod Biol 1994;56:107-10.2. Miller DA,

4. Goodwin TM, Gherman RB, Paul RH. Intrapartum rupture of the unscarred uterus. Obstet Gynecol 1997;89:671-3.

5. Landon MB. Uterine rupture in primigravid women. Obstet Gynecol 2006;108:709-10.

6. Zwart JJ, Richters JM, Ory F, de Vries JI, Bloemenkamp KW, van Roosmalen J. Uterine rupture in The Netherlands: a nation- wide population-based cohort study. BJOG 2009;116:1069–78.

7. Landon MB, Hauth JC, Leveno KJ, Spong CY, Leindecker S, Varner MW, et al.; National Institute of Child Health and Human Development Maternal-Fetal Medicine Units Network. Maternal and perinatal outcomes associated with a trial of labour after prior cesarean delivery. N Engl J Med 2004;351:2581–9.

8. Landon MB, Spong CY, Thom E, Hauth JC, Bloom SL, Varner MW, et al.; National Institute of Child Health and Human Development Maternal-Fetal Medicine Units Network. The risk of uterine rupture with a trial of labour in women with multiple and single prior cesarean delivery. Obstet Gynecol 2006; 108:12–20.

9. Holmgren C, Scott JR, Porter TF, Esplin MS, Bardsley T. Uterine Rupture With Attempted Vaginal Birth After Cesarean Delivery Decision-to-Delivery Time and Neonatal Outcome. Obstet Gynecol 2012; 119: 725–31

10. Kaczmarczyk M, Sparen P, Terry P, Cnattingius S. Risk factors for uterine rupture and neonatal consequences of uterine rupture: a population-based study of successive pregnancies in Sweden. BJOG 2007;114:1208–14.

11. Al-Zirqi I, Stray-Pedersen B, Forsen L, Vangen S. Uterine rupture after previous caesarean section. BJOG 2010;117:809–20

12. Al-Zirqi I, Stray-Pedersen B, Forsen L, Daltveit A, Vangen S. Uterine rupture: trends over 40 years. BJOG 2016;123:780–787.

13. Gyamfi-Bannerman et al. Risk of Uterine Rupture and Placenta Accreta With Prior Uterine Surgery Outside of the Lower Segment. Obstet Gynecol 2012;120:1332–37

14. Bujold E, Jastrow N, Simoneau J, Brunet S, Gauthier RJ. Prediction of complete uterine rupture by sonographic evaluation of the lower uterine segment. Am J Obstet Gynecol 2009; 201: 320.e1–6

15. Kok N, Wiersma IC, Opmeer BC, de Graaf IM, Mol BW, Pajkrt E. Sonographic measurement of lower uterine segment thickness to predict uterine rupture during a trial of labour in women with previous Cesarean section: a meta-analysis. Ultrasound Obstet Gynecol 2014; 42: 132–139.

16. Jastrow N, Chaillet N, Roberge S, Morency AM, Lacasse Y, Bujold E. Sonographic lower uterine segment thickness and risk of uterine scar defect: a systematic review. J Obstet Gynaecol Can 2010; 32: 321–327.

17. Gibbins KJ, Weber T, Holmgren CM, et al. Maternal and fetal morbidity associated with uterine rupture of the unscarred uterus. Am J Obstet Gynecol 2015;213:382.e1-6.

18. Fox et al. Pregnancy Outcomes in Patients With Prior Uterine Rupture or Dehiscence. Obstet Gynecol 2014;123:785–9