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

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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 woman 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.

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
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 95th 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+6 weeks, after adequate metabolic control and daily 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 asymptomatic. She was admitted to the hospital for maternal-fetal surveillance and a new appointment and elective cesarean for 39 weeks were scheduled.

Clinical deterioration: 5th day with antibiotic therapy.

Exploratory laparotomy: vast uterus rupture of posterior wall and a hysterectomy was decided.

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1. 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 |       |       |       |

2. Table 2 Timeline of the patient.

| Dates (in 2016) | 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 | Admission to Maternal-fetal Infirmary for surveillance and therapeutic adjustment with both insulin and metformin |
| 30 August       | 38+2 weeks Asymptomatic      | Adequate metabolic control and daily cardioctography showing fetal well being | Cesarean scheduled to 6 of September |
| 1 September     | Asymptomatic Clinical observation: normal: normal lochia, normal uterine involution and painless examination | Ultrasound on ultrasound | Breast delivery 10 hours later: Dead fetus with 4525 grs |
| 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 metronopen, during three weeks because of multiresistant Klebsiella pneumoniae isolated in the purulent exudate collected from the abdominal cavity during, surgery |

Figure 1: Ultrasound image at emergency department, eight days after delivery (U – uterus; HI – heterogeneous image)
Table 3 Medical History of eight cases of rupture (CS – cesarian; Leuco – leucodermic; melano – melanodermic; leucomelano - leucomelanodermic)

| #  | Race   | Age | Nulliparous | ≥ 3 Deliveries | 1 CS  | ≥ 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             | Yes   | No     | No                                | 3                    | No                   |
| 4  | Leuco  | 44  | Yes (1)     | No             | No    | No     | No                                | -                    | No                   |
| 5  | Melano | 26  | No          | 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                                | -                    | No                   |
| 8  | Leuco  | 37  | No          | No             | No    | No     | No                                | 72                   | No                   |

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 9th 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 > 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)

| #  | Spontaneous\ induction/ cesarian | Time in labor | Timing of diagnosis | Type       | Site of uterine disruption | Other complications | Delivery |
|----|---------------------------------|---------------|---------------------|------------|-----------------|---------------------|----------|
| 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.

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.

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