



Early Challenges: A Case Report of a Premature Baby with Down Syndrome and Uterine Rupture

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Abstract

Introduction Uterine rupture represents one of the most severe obstetric affections. It is defined as a complete or a partial tearing of the uterine wall. Women with a prior cesarean section are reported to have a higher risk of having this situation. Moreover, maternal death and most of all middle- and long-term adverse consequences remain a great preoccupation. On another scale, neonatal death and ulterior deterioration remain very high, especially in low-income countries.

Case Description A 24-year-old woman with a history of previous cesarean section presented at 35 weeks of gestation with pelvic pain without bleeding. Emergency cesarean section revealed a complete uterine rupture at the scar site from the previous cesarean section. Remarkably, the fetus managed to seal the rupture using the right temporal region, forearm, and right leg, avoiding significant complications. The mother had an uncomplicated postoperative course and was discharged after 48 hours of surveillance.

Conclusion We present with this case an extraordinary case of a uterine rupture where both mother and child had a good outcome. This rare evolution was reported only one time in literature. For this reason, a history of caesarean delivery might present a huge challenge for obstetricians and neonates.

Keywords

- ▶ cesarean Section
- ▶ diagnosis
- ▶ perinatal morbidity
- ▶ uterine rupture
- ▶ scarred uterine rupture

Uterine rupture remains a rare but deadly situation, occurring in less than 0.5% of pregnancies worldwide.^{1,2} Higher incidence of uterine rupture cases has been reported in developing countries, especially in African countries, in comparison to developed countries. However, maternal and neonatal death remains notably elevated.^{3,4} In a recent study, the incidence of uterine rupture in the Maternity and Neonatology Center of Bizerte in Tunisia was estimated at 1.7% in women attempting vaginal birth after a previous cesarean section (CS).⁵

Multiple risk factors have been identified in literature as predictive of uterine rupture such as a history of uterine surgery or CS, multiparity, forceps delivery, the inadequate use of uterotonic drugs, and even congenital uterine malformation such as bicornuate uterus.⁶ Review indicated that the majority of uterine rupture occurred in parturient women who have delivered via cesarean in the past.⁷ Signs of uterine rupture vary significantly and are not specific, presenting a diagnostic challenge for practitioners.⁸

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In this article, we describe a case of a uterine rupture in the third trimester with a favorable fetal and maternal outcome.

Case Report

We present a case involving a premature male baby delivered via CS at 35 weeks of gestation. The mother, aged 24, had a medical history of schizophrenia and was under neuroleptic medication. Her previous pregnancy, a year ago, necessitated a CS due to fetal macrosomia and breech position. Prenatal care was limited. Screening for aneuploidies was not performed. Regarding prenatal ultrasound, only the first trimester scan was conducted and no abnormalities were detected.

At 35 weeks, the mother visited the emergency room for pelvic pain without bleeding. An ultrasound revealed severe oligohydramnios, intrauterine growth restriction, bilateral pyelocaliceal dilation, normal-sized kidneys, and duodenal atresia. Fetal distress mandated an emergency CS, during which surgeons discovered a complete uterine rupture at the previous CS scar site.

Remarkably, the fetus managed to seal the rupture using the right temporal region (► Fig. 1A), forearm (► Fig. 1B), and right leg (► Fig. 1C), averting bleeding or membrane rupture. Apgar scores at 1 and 3 minutes were 8 and 10, respectively. The newborn weighed 1,300 g, measured 42 cm in height, and had a head circumference of 28 cm. He was admitted to the neonatal intensive care unit, displaying low muscle tone and Down syndrome's dysmorphic features but no respiratory distress. For the mother, the postoperative course was uncomplicated, and she was discharged home after 48-hour surveillance.

On the 3rd day, surgery for duodenal atresia was successful (duodenoduodenostomy with appendectomy), without complications. At 12 days old, feeding intolerance surfaced,

leading to a 4-day bowel rest, followed by a gradual reintroduction of enteral feeding. Full feeding was achieved at 30 days old.

Subsequent karyotyping confirmed Down syndrome. Echocardiography showed no congenital heart disease. Renal ultrasound revealed minor right-sided pyelocaliceal dilation with no complications. Hypothyroidism was diagnosed on day 11 (thyroid-stimulating hormone >60 mIU/L, free thyroxine = 6.63 pmol/L), prompting initiation of L-thyroxine therapy.

The baby was discharged after 49 days, weighing 2,050 g, and continued L-thyroxine treatment with scheduled outpatient follow-ups.

Discussion

We reported an exceptional instance of uterine rupture where the baby adhered his forehead, left forearm, and leg to the breached area, effectively preventing hemorrhage, umbilical cord prolapse, and amniotic fluid leakage, consequently averting any potential maternal or fetal complications.

Uterine rupture remains one of the most severe incidents for both the baby and the pregnant woman. Studies have shown that prevalence and outcome of uterine rupture widely varies between countries. They are significantly correlated with the national economic, educational, and medical level, and whether women have regular prenatal examination.^{4,9} Scarred uterus, generally a CS scar is still the main risk factor for uterine rupture during pregnancy. In recent years, studies reported more and more uterine rupture secondary to gynecological surgery (uterine fibroids excision accounted for 29.6% of the total number of scarred uterine ruptures, followed by corneal pregnancy resection, 22.2%) with high maternal and neonatal mortality.¹⁰

In our country, authors described a rate of 1.7% of uterine rupture with vaginal birth after CS.⁵ Guidelines have been

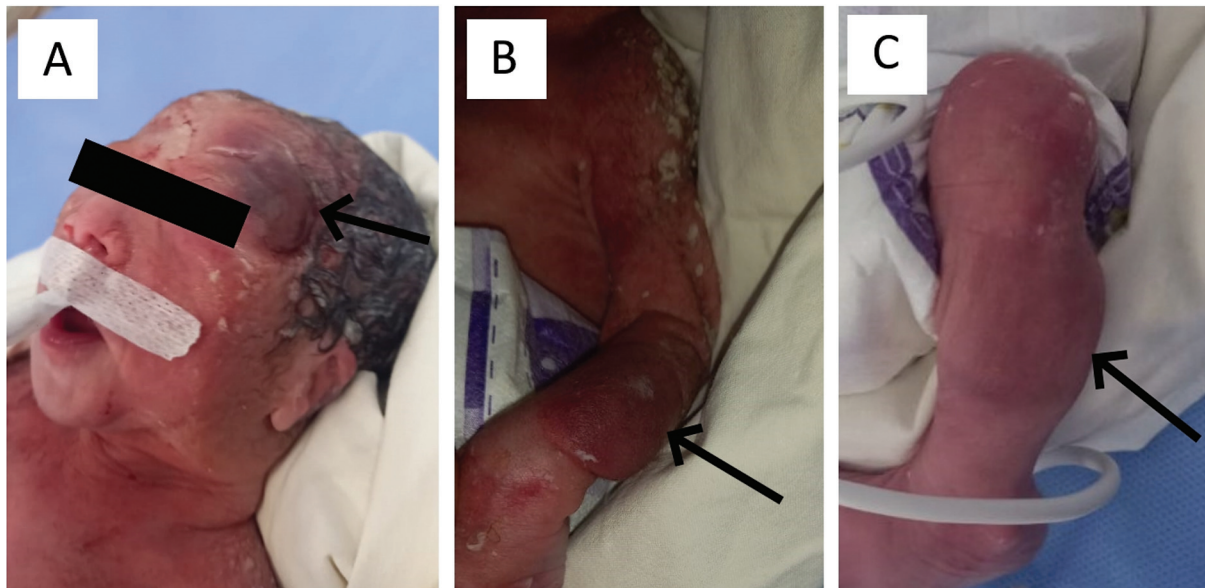


Fig. 1 Temporal (A, black arrows), forearm (B, black arrows), and leg (C, black arrows) hematomas in relation with the closer of the breach of the uterine rupture.

published to avoid this deadly obstetric complication especially in women presented with a previous CS.^{11,12} The risk of uterine rupture is even greater when the previous CS occurred in less than 2 years.¹³

Signs of uterine rupture vary significantly and are not specific, presenting a diagnostic challenge for practitioners. Most common signs are abdominal or pelvic pain, acute vaginal bleeding, changes in the uterine shape with cessation of the contractions, signs of hemodynamic instability, and most of them all fetal heart rate abnormalities.^{6,8}

The described case occurred outside of labor. Besides experiencing abdominal pain, there were no other typical clinical signs of uterine rupture. An incomplete diagnostic picture, notably the absence of contractions, could have contributed to a delay in making the diagnosis. Fortunately, that was not our case.

Both maternal and neonatal mortality remain high.¹⁴ Maternal outcomes are severe, spanning from hypovolemic shock to hysterectomy. On another hand, perinatal consequences remain a serious problem. Savukyne et al reported an overall perinatal mortality of 17%.¹⁵ Other studies described an even higher mortality in unscarred uteri attending 50.6%.¹² Authors have shown that parturients with delayed diagnosis of uterine rupture have poorer maternal outcomes.¹⁴

Regarding fetal prognosis, disruption of oxygen supply to the fetus can cause death or long-term disability. Complete rupture with fetal and/or placental extrusion into the mother's abdominal cavity is associated with worse perinatal outcomes.¹⁴ Davidesko et al showed that several risk factors were identified for intrapartum fetal death, with shoulder dystocia demonstrating the highest risk followed by uterine rupture (adjusted odds ratio [OR] 19.0, 95% confidence interval [CI] 7.0–51.4, $p < 0.05$). Unfortunately, these factors are often neither preventable nor predictable.¹⁶

In a 10-year retrospective study including 27 complete ruptures that occurred among 58614 women, Wu and McGee reported metabolic acidosis among 32% neonates.¹⁷ Within this group, 21% were diagnosed with hypoxic ischemic encephalopathy and two babes (11%) developed cerebral palsy at follow-up. AL-Zirqi et al showed 23.0% neonatal intensive care unit admission, 26.2% neonatal death rate, and 6.1% of ischemic encephalopathy. The authors showed also that the risk of death is significantly increased when the time to delivery was over 30 minutes (OR 16.7; 95% CI 6.4–43.5).¹⁸

Reviewing literature for similar reported cases, only one case has been reported. In 2017, a French team described the case of a 36 weeks' gestation age fetus who saved himself and his mother by being sucked in the uterine tear.¹⁹

Conclusion

The raising rates of cesarean deliveries, especially in developing countries, might present a huge challenge for obstetricians and neonates. Therefore, a better understanding and a better management of this major issue should lead to the establishment of protocols and most of all, the reduction of CS rates.

Declarations

Written informed consent was obtained from both parents.

Consent for Publication

Written informed consent for publication was obtained from both parents.

Availability of Data and Materials

The data sets used during the current study are available from the corresponding author on reasonable request.

Competing Interests

The authors declare that they have no competing interests.

Conflict of Interest

None declared.

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