Uterine Torsion Associated with Preivable Preterm Premature Rupture of Membranes Diagnosed at Cesarean Section

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Abstract

Background: Torsion of gravid uterus is a rare and sporadic event, and its association with preivable preterm premature rupture of membrane (PPROM) is not reported in literature.

Case description: A 35-year-old third gravida with previous two early pregnancy losses was managed conservatively following preivable rupture of membranes at 20 + 6 weeks of gestation. Torsion of gravid uterus to 180° was diagnosed at elective cesarean section performed at 32 + 2 weeks in view of breech presentation with severe oligoamnios and tubular cervix. An alive baby of 1.48 kg with good Apgar score was extracted through a low vertical incision given on anterior surface of uterus after detortion.

Discussion: Torsion is diagnosed when uterus rotates itself on its long axis 45° or more, and the etiology may be fibroid uterus adhesions and ovarian cysts. Preoperative diagnosis is rare except when magnetic resonance imaging is performed. Most of the cases reported in the literature are diagnosed at cesarean section undertaken for fetal distress or nonprogress of labor. In the case presented here, there were no predisposing factors except that she was on prolonged bed rest due to PPROM, and there was a cornual implantation of placenta.

Conclusion: Conservative management of maternal position may predispose to uterine torsion, and in such cases, vigilant monitoring and timely termination can save the fetus.

Clinical significance: Presence of a tubular cervix with mal-presentation is one of the clinical features to suspect torsion during pregnancy and labor.

Keywords: Breech presentation, Preivable PPROM, Recurrent pregnancy loss, Torsion gravid uterus, Tubular cervix.


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Background

Torsion of gravid uterus is a sporadic event and is associated with high perinatal mortality. It is an obstetric surprise and is usually recognized during cesarean section either before the delivery of the baby or after delivery. A review over a period of 40 years (1966–2006) could find only 38 cases.¹ The predisposing factors or causes are not clear, and encountering such an event warrants reporting. The association of preivable PPROM with torsion uterus is also not reported in the literature. Here we present a woman of recurrent pregnancy loss with preivable preterm PROM in whom torsion was encountered at cesarean section.

Case Description

A 35-year-old G3 A2 presented to the emergency department at 20 + 6 days of gestation with history of leaking per vaginum of 8 hours duration. This pregnancy resulted after undergoing an intrauterine insemination for secondary infertility. Her first pregnancy was spontaneous conception in 2013, and it ended as incomplete abortion at 8 weeks for which she underwent dilatation and curettage. The second pregnancy was after 2 years following ovulation induction and ended as missed abortion at 9 + 5 weeks and was again treated with dilatation and curettage. She was diagnosed as GDM during the second pregnancy at the time of missed abortion and was treated with metformin. Subsequently she has not followed up for screening of type 2 diabetes mellitus.

During the current pregnancy, she underwent first-trimester biochemical screening and was told to be at high risk for Down syndrome elsewhere (FTS 1:12). She consulted at our hospital at 13 + 6 weeks, and NT scan was normal and nasal bone was seen; hence, she was reassured and was advised to undergo GTT along with other antenatal screening tests. After that she did not come for her regular visit but presented only at 20 + 6 weeks with leaking. She was hospitalized as there was demonstrable leaking of liquor and on USG liquor was less with SDP of 1.8 cm. Biometry corresponded to 18 + 4 weeks with good cardiac activity. She was treated with bed rest, foot end elevation, oral hydration, tocolytics, and injection 17 hydroxyprogesterone caproate 500 mg intramuscularly weekly. Her cervical swab grew Enterococcus spp., and she was treated with sensitive antibiotics for 1 week. She was diagnosed as GDM and was on diabetic diet. As the patient and relatives were anxious about the fetal anomaly, a second opinion from radiologist was obtained. Renal agenesis was suspected, as they could not visualize the kidneys. She was treated with intravenous fluids on alternate day in addition to oral hydration and left lateral position. She was soaking one to two pats per day and recurrence of cervicovaginal infection.
Hence, she was advised metformin along with diet, and her blood sugar profile was monitored every 2 weeks, and she received a second course of antibiotics. Repeat ultrasound evaluation after 17 days of admission showed bilateral normal kidneys and good diastolic blood flow in umbilical artery and a liquor pocket of 2 cm. This was confirmed by a third opinion from Fetal Medicine Center (Mediscan, Chennai), and the patient and her relatives were reassured. Conservative management was continued, and she was monitored weekly with USG. Liquor improved gradually to 6 cm, and she stopped leaking at 28 weeks. Antenatal corticosteroids were administered under the cover of buffered dextrose. She was planned to discharge at 30 weeks as AFI was 8 cm. But she suddenly leaked the next day of decision to discharge and AFI was 2 cm. Cervical swab culture grew *Escherichia coli* and white blood cell count was 15,000/mm³. She was treated conservatively further, and liquor did not improve, and at 32 + 2 weeks, the USG evaluation showed no cord free pocket liquor and the fetus was in transverse position with an estimated weight of 1.4 kg with PI of 0.8. Placenta was posterior type IIB. Cervix appeared tubular. A decision was made to deliver by elective cesarean section after discussion with the neonatologist explaining the risks and benefits. At cesarean section, on opening the peritoneum, the fallopian tube and a congested ovary were present at the incision site, and the gravid uterus was found to be torted to 180° in anticlock-wise direction (Fig. 1). Detortion was done (Fig. 2), and a vertical incision was given on the lower segment with 2-cm extension to upper segment, and the fetus was gently extracted as breech. Alive, female fetus with APGAR 8/10 at 5 minutes weighed 1.48 kg, and there were no congenital anomalies. The cord and membranes showed velamentous insertion (Fig. 3). The placenta was adherent and implanted cornually and posteriorly and needed manual removal (Fig. 4), as there were no signs of separation even after waiting for 15 min. Both ovaries were polycystic, and there were no adhesions or uterine malformations. The uterine incision was closed in two layers (Fig. 5) The abdomen was closed in layers. The postoperative course was normal. The baby was neonatal intensive care unit (NICU) for a period of 6 weeks (42 days) and was discharged home healthy and weighed 1.9 kg. The total duration of hospital stay was 120 days.

**DISCUSSION**

Managing a pregnant woman with previable preterm premature rupture of membrane (PPROM) as early as 16 weeks is a challenging and controversial issue in obstetrics. When a women with recurrent pregnancy loss and without a live baby approaches for management, conservative approach is to be advocated although her amniotic fluid is continuously leaking.
Torsion of Gravid Uterus and Pregnancy Outcome

Torsion is diagnosed when the uterus rotates on its long axis by 45° or more at cervicoisthmical junction. Affects of torsion of gravid uterus range from asymptomatic status to complete gangrene and depend on the degree of rotation and duration of gestation and the etiology. The etiology includes fibroid uterus in 30%, uterine anomaly in 15%, and pelvic adhesions and ovarian cysts (15%); the cause is unknown in the remaining. Maternal position, posture, and irregular body movements were supposed to be the triggering factors for torsion.\(^2\) In this case, the lady kept herself in left lateral position with support almost all the time.

The symptoms arising due to torsion are nonspecific, and the reported symptoms are acute abdominal pain, nonprogress of labor, and suspicious and pathological fetal heart pattern.\(^3,4\) The affects of torsion depend up on the degree of torsion, and the most commonly reported degree of torsion is 90°–180°. A torsion of 540° was reported to be associated with uterine necrosis, and an extreme torsion of 720° with intrauterine death was reported due to cord-like fibrosis of cervix which was misdiagnosed a cervical stenosis on MRI.\(^2\)

Preoperative diagnosis is not made usually, but in the recent times, it was possible after subjecting to magnetic resonance imaging (MRI) with high degree of clinical suspicion. The first reported case of uterine torsion by MRI was in 1995\(^5\) although the first case of torsion was published in 1876 by Labbé. In a review involving 38 cases only, 1 case was reported to be diagnosed prior to delivery by MRI.\(^1\) Vagina is normally seen as H-shaped structure on MRI but appears as X when torsion occurs.

In most of the cases reported at term, the torsion was recognized after delivery of the fetus through the incision was on posterior wall.\(^3,4\) This mostly occurred in torsion of 180° with a live fetus taken up for cesarean for fetal distress. In cases of torsion of 45°–90°, it is easy to recognize and detort prior to incision. In this case, torsion occurred up to 180° in anticlockwise direction and was easy to detort due to lack of adhesions or tumors. Because of the anticipation of tubular lower segment due to preterm, a low vertical incision was planned preoperatively. Cornual implantation of placenta and maternal position may be the predisposing factors for torsion in this case. High maternal and perinatal mortality was reported when torsion occurs more than 180°.\(^6\) Maternal mortality is rarely reported in modern obstetrics due to this condition, but still perinatal mortality is approximately 18%.\(^1\) Early and timely decision to deliver the fetus by cesarean section is necessary to avoid the morbidity and mortality associated with the condition, and a high index of suspicion is necessary to diagnose the condition preoperatively.

CONCLUSION

Conservative management of previable PPROM may be successful in selected cases and should be practiced especially in women with RPL. Maternal position may predispose to uterine torsion in such cases, and vigilant monitoring and timely termination can save the fetus.

CLINICAL SIGNIFICANCE

- Decision-making in conservative management of previable PPROM is a dilemma. No clinical guidelines exist.
- Termination of pregnancy in such cases is to be undertaken after taking in to account the gestational age, fetal survival, and availability of advanced NICU care. Mode of delivery should be least traumatic to the fetus.
- High index of suspicion is essential to diagnose uterine torsion during pregnancy. Presence of a tubular cervix with mal-presentation is one of the suspecting clinical feature of torsion.

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REFERENCES