

DELAYED-INTERVAL DELIVERY WITH TIMELY CORD LIGATION

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A 43-year-old gravida 6 para woman with dichorionic diamniotic (DCDA) twins presented at 18+6 weeks gestation with extreme Preterm Premature Rupture of Membranes (PPROM) of twin 2. 23 days later at gestation 22+1 the patient had cord prolapse of twin 1 with an absent fetal heart. The cord of twin 1 was ligated under general anaesthetic with subsequent spontaneous vaginal delivery of twin 1. The patient was kept as an inpatient and monitored for signs of chorioamnionitis and was continued on an antibiotic regimen and steroid loaded. At 31+4 twin 2 was delivered by emergency caesarean section in good condition, giving an interval delivery of 63 days. In this case we believe that the counselling and management of the cord prolapse with high ligation along with close monitoring reduced the risk of infection and resulted in delaying the delivery of twin 2, resulting in an excellent outcome for both mother and baby.

KEYWORDS Interval Delivery, Cord Ligation, Twin Pregnancy		
Article History		
Received	Accepted	Published
23/05/2018	06/07/2018	05/12/2018
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CASE REPORT

A 43-year-old gravida 6 para 2 abortus 3 woman with dichorionic diamniotic (DCDA) twins presented at 18+6 weeks gestation to the Royal Women's Hospital in Melbourne, Australia with extreme Preterm Premature Rupture of Membranes (PPROM) of twin 1. Her obstetric history included two normal vaginal deliveries 26 and 24 years prior and 3 previous In-Vitro Fertilisation (IVF) cycles resulting in miscarriages. The current pregnancy was with a new partner and was the result of double embryo transfer. The antenatal course has been unremarkable with Non Invasive Prenatal Testing (NIPT) showing low risk for aneuploidy. On admission rapid immunoassay (AmniSure ROM test) confirmed PPROM and the patient was admitted for intravenous amoxicillin for 48 hours and commenced on oral erythromycin. A guarded prognosis was given to the patient who elected for expectant management and for monitoring on the antenatal ward.

23 days later at gestation 22+1 the patient was transferred to birth suite with a cord prolapse of twin 1. Real time scan showed Twin 1 was transverse and Twin 2 was breech and both had fetal heart rates (FHR). Speculum showed a long but slightly open cervix with a loop of cord at introitus. The patient requested conservative management and the cord were pushed back into the uterus with no success. Following this a FHR for twin 1 was unable to be found, and Fetal Death in Utero (FDIU) was confirmed. At 22+2 the cord of twin 1 was ligated under general anaesthetic to reduce the risk of infection. Intraoperatively there was 20 cm of cord outside of the vagina and the cord with ligated at cervix and tied off with PDS 0. The cord stump was placed inside cervix, which was 1.5cm dilated and 2cm long. At 22+4 the patient went into spontaneous labour and delivered a stillborn female weighing 380g with placenta remaining in situ. Contractions ceased with no signs of impeding delivery of twin 2. Histopathology of the ligated cord showed features consistent with severe acute funisitis (stage 3).

The patient was kept as an inpatient and monitored for signs of chorioamnionitis using Full Blood Count, High Vaginal Swabs and C-Reactive Protein (CRP) and was continued on the above antibiotics until 23+3 where she was changed to oral amoxicillin, with all antibiotics being ceased at 24+4. Betamethasone was administered at 22+5 and 22+6 with booster steroids given weekly for two subsequent weeks. At 29+5 the fetus was biophysically well with an Estimated Fetal Weight on the 10th centile and the patient was discharged home. She was readmitted at 31+1 for a small Antepartum Haemorrhage with speculum exam showing a 3cm dilated cervix and bulging membranes. A further dose of steroids was administered and she was subsequently induced at 31+4 due to risk of infection as membranes were at the

introitus.

At 31+4 twin 2 was delivered by emergency caesarean section due to suspected placental abruption giving an interval delivery of 63 days. A female neonate was delivered in good condition with APGARS of 9, 9 and a weight of 1466g. There was 100mL of clot in the uterus, and twin 1 placenta was peeled with difficulty. The neonate required time in Neonatal Intensive Care Unit for prematurity, suspected sepsis and hypoglycaemia and was discharged to a satellite special care nursery on day 14 and discharged home day 47.

DISCUSSION

Twin pregnancies are six times more likely to result in a preterm delivery (<37 weeks) compared to singletons, with 50% of all twins being born premature. The prevalence of neurodevelopmental disability is consistently higher in twins than in singletons due to both birth weight and gestational age being lower 2 An infant born at 22 to 25 weeks gestation has a mortality rate of greater than 50%, and nearly half of the surviving infants are left with significant neurodevelopmental disabilities 3

Whilst most twin pregnancies involve the birth of the first twin followed by the second twin, delayed-interval delivery involves a longer period after the birth of the first twin in order for the second twin to achieve viability and have improved survival and neurodevelopmental outcomes. A systematic review looking at 128 cases of interval delivery found that second born had a significantly lower mortality risk compared to the first born.4 Whilst there are many case reports in the literature describing delayed-interval delivery, the is a lack of randomised controlled trials due to the rarity of this condition means management is patient specific and variable.5,6 This patient was a good candidate for interval delivery as it was a DCDA pregnancy with the membranes of the second fetus remaining intact with no evidence of infection or fetal anomalies. The patient and partner were counselled about the risks of preterm delivery and maternal infection and wished proceed with delayed-interval delivery of twin 2.

There are many cases of cord ligation of twin 1 after delivery in order to reduce the risk of ascending infection from the vagina involved in delayed-interval delivery.6-9 Previously at our hospital there was a case of interval delivery after PPROM at 18 weeks gestation with cord ligation after delivery of twin 1, and delivery of twin 2 46 days later at 25+4 days.8 However all these cases involved cord ligation after delivery of twin 1 as the placenta was still in utero. This case is unique as it involved cord prolapse and subsequent FDIU of twin 1 which was then followed by cord ligation. Only after did the patient go into spontaneous labour and deliver twin 1. Rather than waiting for delivery of twin 1 after PPROM, the decision to ligate and tie off the

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cord potentially reduced the risk of infection along with the use of broad-spectrum antibiotics.

One study found that placing a cervical cerclage after the first delivery was associated with a longer delivery interval and higher birthweight of the second fetus.10 In this case, as the cervix closed after the delivery of the first twin, the decision was to not place a cerclage. Similarly the quiescence of contractions negated the use of tocolytics, which according to the literature can be used to prevent delivery of second twin in presence of uterine activity.5,11 The use of steroids at the cusp of viability was in line with current literature, as an intramuscular course of betamethasone or dexamethasone helps pulmonary epithelial development and matures other organ systems.12 The enabled us to offer resuscitation at 22+6 weeks.

Although delayed-interval delivery may improve neonatal mortality and morbidity, maternal morbidity must also be considered. One study found that 31.6% of patients experience serious maternal morbidity, the main risks being intrauterine infection and sepsis.5,11 In this case there was no maternal infection, likely due to the combination prophylactic antibiotics as well as close monitoring of blood work and high vaginal swabs.

While there have been multiple cases on interval delivery in the literature, to our knowledge this is the first instance where the cord was ligated prior to delivery of twin 1. Delayed-interval delivery can result in the delivery of the second twin at a viable gestation with better neurodevelopmental outcomes, however the maternal risk must also be weighed. Each case must be assessed independently while further research must be done in order to standardise treatment. In this case we believe that the counselling and management of the cord prolapse with high ligation along with close monitoring reduced the risk of infection and resulted in delaying the delivery of twin 2, resulting in an excellent outcome for both mother and baby.

CONFLICTS OF INTEREST

The author declares that there is no conflict of interest regarding the publication of this paper.

CONSENT

Written consent to prepare this manuscript was given by the patient.

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