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Delayed-Interval Delivery of Twin Gestation via Cesarean Section: A Case Report

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Study Design A
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Patient: Female, 21
Final Diagnosis: Preterm prelabor rupture of membrane
Symptoms: Cord prolapse • rupture of membranes
Medication: —
Clinical Procedure: Cesarean section
Specialty: Obstetrics and Gynecology

Objective: Unusual clinical course
Background: Multifetal pregnancies are at high risk for preterm delivery. Under certain circumstances, delayed vaginal delivery of the second twin is performed to improve morbidity and mortality. Most of the information on optimal management of delayed-interval delivery comes from published case reports in which the first twin was delivered vaginally. This case report is unique in that twin A was delivered via cesarean section.

Case Report: Our patient was a 21-year-old G2P1, with dichorionic diamniotic twins of unknown gestational age, with prenatal care at a different facility, who presented with preterm prelabor rupture of membranes and cord prolapse. Twin A, with an estimated weight by ultrasound of 528 g, was delivered via cesarean section and twin B was left in utero until the patient went into preterm labor 10 days later. Obstetrical management included tocolytic protocol from the Management of Myelomeningocele Study trial, preterm prelabor rupture of membrane antibiotics with broad-spectrum coverage, and judicious use of fetal lung maturity steroids and magnesium sulfate.

Conclusions: This case is important as we have demonstrated that cesarean section in the setting of delayed-interval delivery may be an option to improve survival at the limits of viability. We also discussed our treatment approach and how we delayed delivery of the second twin by 10 days. Unexpectedly, the surviving twin was the one born first, at 22 4/7 weeks determined 2 days after birth by prenatal records.

MeSH Keywords: Cesarean Section • Fetal Membranes, Premature Rupture • Obstetric Labor, Premature • Pregnancy, Multiple

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Background

Multifetal pregnancies are at high risk for preterm delivery. In most pregnancies, the delivery of the first twin is immediately followed by delivery of the second twin. However, under certain clinical circumstances, delayed delivery of the second twin has been reported [1–14]. The interval range is wide (1–152 days) [1], the length being inversely proportional to the gestational age at which the first twin was delivered [2]. Ideal candidates for delayed-interval delivery of the second twin are those who spontaneously deliver a fetus at the limits of viability, due to cervical insufficiency, preterm labor or premature rupture of membranes, or intrauterine demise [2]. Delayed delivery for the second twin until a later gestational age may help to improve morbidity and mortality, but risks of chorioamnionitis, placental abruption, preterm labor, and premature rupture of the membranes are elevated [8,10]. This complex clinical scenario is best handled by a multidisciplinary team consisting of obstetricians, maternal fetal medicine consultants, neonatologists, and the patient.

Most of the information on optimal management of delayed-interval or asynchronous delivery of twins comes from published case reports in which the first twin was delivered vaginally. This case report is unique in that twin A was delivered via cesarean section. To the best of our knowledge, there are no previous reports of delayed-interval delivery with the first twin delivered by cesarean section.

Case Report

Our patient was a 21-year-old G2P1, who presented to labor and delivery with a twin pregnancy of unknown gestational age, with prenatal care at a different health care system. She was complaining of loss of fluid and something “falling out of the vagina.” Her past medical history consisted only of herpes simplex virus, but she denied any active lesions or prodromal symptoms. Her obstetrical history was noted for a full-term uncomplicated vaginal delivery 2 years prior. Speculum exam revealed gross pooling, cervical dilation of 2 cm, and a prolapsed umbilical cord in the vaginal vault. No prenatal records were available at the time. A bedside ultrasound demonstrated dichorionic diamniotic twins in cephalic presentation with normal fetal heart rates in both. Twin B was noted to have an intact amniotic sac with normal fluid levels. The estimated gestational weight for twin A was 528 g and 568 g for twin B, and both had estimated gestational ages above 23 weeks. She was admitted for preterm prelabor rupture of membranes (PPROM) of twin A complicated by a prolapsed umbilical cord.

The patient and her family were extensively and expediently counseled regarding the diagnosis and the poor prognosis for

both fetuses if delivered due to the peri-viable gestational age. Our internal protocol is to offer neonatal resuscitative efforts at a gestational age >23 weeks or estimated fetal weight >500 g. The option for delayed-interval delivery suggested, if feasible, to provide a better prognosis for the second fetus. An option of induction of labor to deliver vaginally was discussed, with the clear likelihood of both fetuses being delivered. Expectant management was also reviewed, and secondary to strong personal beliefs, the patient and family wanted heroic measures for both babies, regardless of the poor prognosis and limits of resuscitative efforts at such an early gestational age. Following a multidisciplinary family conference, the patient elected to proceed with cesarean delivery with attempted delayed-interval delivery of the second twin. Approximately 2 h elapsed between presenting to the hospital and delivery of the first twin, with most of the time spent on counseling. Surprisingly, the fetal heart rate of twin A remained normal for gestational age despite the prolapsed umbilical cord.

A single dose of fetal lung maturity steroids and cefoxitin was given prior to skin incision. A low vertical uterine cesarean section was performed with an estimated blood loss of 500 g. Twin A, a male, was delivered in cephalic presentation and the umbilical cord was suture-ligated with 0-vicryl as close to the insertion site as possible. Apgar scores were 1, 7, and 8 at 1, 5, and 10 min, respectively and weight was 530 g. The infant was intubated and taken to the neonatal intensive care unit (NICU) for stabilization. The amniotic sac for twin B was noted to be intact and the uterine incision was closed with 0-vicryl in a running fashion. Terbutaline 0.2 mcg was given intraoperatively. The patient was transferred to recovery in stable condition. Fetal heart tones for twin B were obtained and the patient was transferred back to labor and delivery for further management. Prenatal records obtained 2 days later confirmed dichorionic diamniotic twins and dated the pregnancy at 22 4/7 weeks by a 10-week ultrasound.

On postoperative day (POD) #1, patient had stable vital signs and denied any contractions. She endorsed fetal movement and some vaginal spotting. She was placed on continuous fetal monitoring. The patient received a second dose of fetal lung maturity steroids and received 2 additional doses of cefoxitin. Her post-operative WBC was $18.3 \times 10^9/L$ and her Hb was 9.5 g/dL from a baseline of $13.5 \times 10^9/L$, and 12.1 g/dL. We elected to follow the post-operative treatment plan used following hysterotomy in the Management of Myelomeningocele Study (MOMS) trial, and started indomethacin 50 mg PO Q6h for the first 24 h and 25 mg PO Q6h for an additional 24 h [15]. Nifedipine 20 mg PO Q6h was then started for maintenance. Latency antibiotics for PPRM were also administered (ampicillin 2 g IV Q8h and erythromycin 250 mg IV Q8h). Because this patient had undergone surgery in the setting of PPRM and cervical dilation, we decided to include clindamycin to

her antibiotic regimen to broaden the antimicrobial coverage (clindamycin 900 mg IV Q6h). Complete blood count and C-reactive protein were ordered Q72h. Cerclage was considered, but deferred due to persistent vaginal spotting. The goal was delivery of twin B by repeat cesarean section at 34 weeks or earlier for maternal or fetal indication.

On POD #2, she remained stable and again denied any contractions, but continued to have minimum vaginal spotting. She transitioned to intermittent monitoring Q4h. The patient continued her intravenous antibiotic regimen and indomethacin. Her Hb was noted to be 7.5 g/dL, likely due to a combination of blood loss from surgery and continued vaginal bleeding postoperatively. However, the patient was asymptomatic and the fetal heart tracing remained category I. The patient declined a blood transfusion and intravenous iron was started. Serial daily ultrasound showed no evidence of abruption. On POD #3, the patient remained stable and she completed her intravenous antibiotics and started oral antibiotics. She continued nifedipine 20 mg PO Q6h for tocolysis. On POD #4, the clindamycin was changed to 600 mg PO Q12h. On POD #5, her Hb had dropped to 7.2 g/dL. She was transfused with 2 units of packed red blood cells. The post-transfusion Hb was 10.9 g/dL. A rescue dose of fetal lung maturity steroids was given on POD #8, 7 days after the initial course. Our protocol is to administer a rescue dose 7 days after the prior dose if delivery appears imminent and the delivery is at the limits of viability, as the benefits of steroids for neonatal risk reduction wane after 1 week [16,17]. Given that this patient had multiple risks factors for early delivery of twin B, we elected to administer a rescue dose on POD #8. At this time, she had finished her antibiotics and she was continued on her tocolytic regimen with occasional doses held due to low blood pressure.

On POD #9, the patient complained of increased vaginal bleeding. Her vitals were stable and the fetal heart tracing was reassuring. Her vaginal bleeding was monitored closely throughout the day and was noted to be minimal. Late that night, she reported passing a large blood clot when she ambulated to the restroom and continuous fetal monitoring was resumed. Fetal heart tracing noted moderate variability with variable decelerations noted about every 10 min. Resuscitation efforts were performed due to the category II tracing. Early on POD#10, a speculum exam was performed for continued vaginal bleeding; a blood clot was noted in the vaginal canal and her cervix was now 3 cm dilated. Magnesium sulfate was started for neuroprotection. She quickly progressed to 4–5 cm cervical dilation, had increased vaginal bleeding, and a category II tracing that was unresponsive to resuscitation efforts. She was taken to the operating room for an emergent repeat cesarean section. Twin B, a female, was delivered at 24 0/7 weeks of gestation using the same low vertical hysterotomy incision. The infant weighed 600 g and had Apgar scores of 1

and 8 at 1 and 5 mins. The infant was intubated in the operating room and transferred to the NICU.

The maternal post-partum course was complicated by endometritis, for which she was treated with antibiotics, and she was safely discharged home on postoperative day 3. Twin A remained in the NICU for approximately 4.5 months and then was discharged home. Twin B died on hospital day 2 due to cardiovascular and respiratory failure complicated by tension pneumothorax and hypotension that was refractory to medication. A follow-up exam of twin A demonstrated a diagnosis of chronic lung disease of prematurity, retinopathy of prematurity requiring laser photocoagulation, and seizures that resolved by 1 year of age. At 18 months, the infant was meeting motor milestones but was speech-delayed.

This patient subsequently conceived within 3 months of her twin delivery. She was followed closely in our high-risk clinic, but had spontaneous preterm labor and delivered via repeat cesarean section at 29 5/7 weeks of gestation.

Discussion

There are several aspects of this case that make it unique. The most obvious is that a cesarean section was performed to deliver both fetuses. It is also notable that this patient was young and spontaneously conceived twins. The majority of previously reported delayed-interval deliveries involved pregnancies conceived through *in vitro* fertilization in an older population. She also received a blood transfusion between the 2 surgeries. The anemia could theoretically lead to fetal hypoxia in twin B. Twin A received a single dose of fetal lung maturity steroids just prior to delivery and twin B received both doses and a booster 1 week later. It is unclear how these factors contributed to the success or failure of maternal and neonatal outcomes.

There is a lack of consensus regarding optimal obstetrical management in rare situations of delayed-interval or asynchronous twin delivery. Management recommendations are extrapolated from isolated case reports and include tocolytics, broad-spectrum antibiotics, amniocentesis of the second fetus, fetal lung maturity steroids, and placement of a cervical cerclage [1,3,9,13]. Some authors report that cerclage leads to a longer delayed delivery interval and some argue that cerclage increases the risk for infection and rupture of membranes [3]. Other recommendations include vaginal washes with chlorhexidine, cervico-vaginal cultures, monitoring of infectious laboratory parameters such as WBC and C-reactive protein, and vaginal progesterone [3,4,14].

Proposed inclusion criteria for a delayed-interval delivery of a second twin are: birth of the first fetus typically around

peri-viability (some authors suggest up to 30 weeks of gestation), diamniotic pregnancy, intact membranes in the second twin, absence of intrauterine infection, and absence of fetal or maternal pathology that requires urgent termination of pregnancy [5]. Neonatal morbidity is primarily dependent on gestational age at the time of delivery. A systematic review of the survival benefit of the second twin from delayed-interval delivery demonstrated significantly lower mortality for the second fetus compared to the first [6]. Delayed-interval delivery is associated with maternal morbidity, with the most common being intrauterine infection, occurring in 17–52% of cases [7]. The most common reason for delivery of the second fetus is preterm labor (with or without rupture membranes) and clinical evidence of intrauterine infection [2].

We present this case to demonstrate that delayed-interval delivery with cesarean delivery of the first twin is possible. The use of the MOMS tocolytic protocol following delivery seems reasonable and appears to have aided the progression of the pregnancy by another 10 days between the 2 cesarean sections. It seems reasonable to use this regimen in similar situations in the future. Clindamycin was also given in addition to PPRM antibiotics due to the patient's high risk for infection. Although the placenta demonstrated evidence of chorioamnionitis, it is unclear whether this occurred prior to rupture of membranes (and contributed to the PPRM) or between the 2 deliveries (and contributed to the preterm delivery of twin B).

While most delayed-interval deliveries are done to improve survival of the second fetus, this did not occur in our case. Twin A survived and twin B died. Twin A survived despite being delivered at an earlier gestation, being male, and not being exposed to maternal corticosteroid injection. Twin B died due to complications from a tension pneumothorax and hypotension, which could have been due to respiratory distress syndrome, mechanical ventilation, or extreme prematurity [18]. While we will never know if twin B would have survived if we had delivered it at the same time as twin A, at the time it was a reasonable decision to delay delivery to provide a better prognosis for twin B. It is also unclear if twin B benefited from any additional measures given, such as steroids or magnesium sulfate, or was harmed by the patient's post-operative anemia and transfusion. Neonatal mortality and morbidity are high in the peri-viable period, regardless of the circumstances.

Conclusions

There are several large reports on delayed-interval delivery of multifetal gestations, in which the first fetus is delivered vaginally. We know of no reported cases in which the first fetus was delivered surgically. This case is important because we demonstrated that cesarean section in the setting of delayed-interval delivery may be an option to improve survival at the limits of viability. We also discussed our treatment approach and how we delayed delivery of the second twin by 10 days.

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