Prolongation of Second Twin's Delivery Until Term: A Rare Case of Delayed-Interval Delivery

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Although some cases describing delayed birth of the second twin have been published recently, delay of delivery beyond 36 weeks seems scarce. We report a case of delayed-interval delivery wherein prolongation of the second twin's delivery until term with a subsequent favorable infantile outcome. In this case, the stillbirth of the first twin occurred at 25 weeks' gestation. Prophylactic tocolysis was performed with ritodrine and magnesium sulfate, and a McDonald cerclage was performed 2 days after delivery of the first twin. Ampicillin and gentamicin were also administered for the purpose of prevention of intrauterine infection. No clinical sign of chorioamnionitis was found thereafter, and full term uneventful delivery was achieved. With this experience, we believe that delayed-interval delivery can be effective in prolonging gestation and should be an option if the first twin developed an extreme preterm delivery.

Keywords: Cervical cerclage; Delayed delivery; Diamniotic dichorionic twin

In twin pregnancies, there is a high risk of preterm delivery, resulting in infant morbidity and mortality. Gestational age is an important predictor of neonatal survival; therefore, strenuous efforts have continued worldwide to avoid preterm labor. If the first fetus is vaginally delivered during the extremely preterm period, the delivery of the second twin generally follows shortly thereafter. However, some cases with an outcome of the delayed birth of the second twin have been recently published reporting favorable outcomes. Herein, we report a rare case of delayed interval delivery wherein prolongation of the second twin's delivery until term was achieved after the stillbirth of the first twin at 25 weeks' gestation.

Case Presentation
A woman (gravida 3, para 1), aged 37 years, with dichorionic diamniotic twin pregnancy after spontaneous ovulation was referred to our hospital at 19 weeks and 6 days because of fetal pleural effusion of the first twin. On referral, ultrasonography examination revealed cystic hygroma of the neck, pleural effusion, and systemic subcutaneous edema in the first twin. No anomaly was noted in the second twin. Two independent placetas were visualized. Chromosomal analysis via amniocentesis was not performed prenatally owing to lack of patient consent. During subsequent follow-up, the first twin developed to intrauterine death at 23 weeks and 4 days. No evidence of maternal mirror syndrome was observed throughout the pregnancy. At 25 weeks and 4 days of gestation, she experienced onset of uterine contractility with premature uterine full dilatation and admitted. No clinical sign of chorioamnionitis was found. Labor pain was not exhibited, and the stillbirth of the first twin occurred 1 hour after the admission. Usually, cesarean sections are performed in cases of extremely preterm twin births; however, the first twin was rapidly delivered vaginally. Therefore, delayed delivery was an alternative. After the delivery of the first twin, the options available were either delayed interval delivery or immediate oxytocin augmentation to induce delivery of the second twin. The patient and her family were informed about the option for delaying the remaining twin and the possible following...
complications. They were in agreement, and therefore, we decided to retain the second twin to allow improvement in the outcome. The umbilical cord of the first twin was ligated with an absorbable suture as high in the cervix as possible, and returned to the uterine cavity. The placenta was left inside the uterus. Prophylactic tocolysis was performed with ritodrine and magnesium sulfate. Intravenous ampicillin (6 mg per day) and intravenous gentamicin (240 mg per day) were administered for 7 days to prevent intrauterine infection. Betamethasone (12 mg) was administered intramuscularly twice a day to promote the maturity of the second twin. There was no clinical sign of chorioamnionitis, and we performed a McDonald cerclage at 25 weeks and 6 days (2 days after delivery of the first twin). Cervical length was sonographically followed up twice a week, and did not show remarkable progress. Uterine contraction was exhibited, and tocolytic agents were discontinued at 32 weeks of gestation. After that, the pregnancy continued without enhancement of uterine contraction, and ultrasound examination showed normal growth and cephalic position of the second twin. As the mother showed no signs of infection according to physical (heart rate and body temperature) and laboratory (blood cell counts and C-reactive protein level) findings, she was discharged from our hospital at 36 weeks’ gestation (Figure 1). Cervical cerclage suture was removed on that day. At 37 weeks and 5 days of gestation, spontaneous labor pains occurred, and she was admitted again. On that same day, labor progressed uneventfully, and a male infant weighing 2278 g was born with an Apgar score of 9 to 9. The delivery interval between the twins was 83 days. The placenta of the dead twin was atrophic (not hydropic) in comparison with placenta of the viable twin (Figure 2). We were unable to obtain patient consent (from the pregnant woman) to perform chromosomal microarray analysis of the delivered placenta. A piece of placenta of the viable twin was investigated for histological changes, which revealed that fetal membrane and umbilical cord had no specific inflammation. Follow-up at 6 months of life demonstrated uneventful development.

Discussion
It is well known that twin pregnancies are associated with a higher risk of preterm delivery, and at a significantly earlier gestational age than singletons, resulting in infant morbidity and mortality. 1 Delivery of the presenting fetus in twin gestation is usually followed by delivery of the second fetus within a short period of time. Traditionally, this situation is managed by delivery of both fetuses, either vaginally or by cesarean section. However, gestational age is the most
important predictor of neonatal survival in immature infants. Especially, extreme prematurity – before 25 weeks of gestation – is associated with poor postnatal outcome. Prolongation of gestational period and increase in fetal weight significantly improve the outcome. Considering these, in recent years, an attempt to prolong the pregnancy for the second twin, even after the first twin’s delivery, has increasingly been discussed, and a large number of cases with good results have been published. According to a prospective study published by Arabin and van Eyck with obstetric follow-up of 50 cases of multiple pregnancies, the delayed birth of the second twin is associated with better perinatal results if the birth of the first twin happens between weeks 20 and 29.

The reported intervals varied widely. In a population-based study that included 200 pregnancies with delayed delivery, Zhang et al reported that the median duration of delay was 6 days. An average interval delivery of 6.33 days (1-14 days) was also reported in a case series by Benito Vielba et al. Since delayed-interval delivery is an attempt applied during the extreme preterm period, prolonging a pregnancy by 6 days must contribute to improvement of the infantile outcome. Several investigators have reported longer intervals. Rosbergen et al published a study of 24 cases of attempted delayed-interval delivery and described that the mean of the delay was 19.9 days. Kalchbrenner et al published a study of seven cases and reported delayed delivery of the second twin with a mean of delivery interval of 36.2 days. Fayad et al reported a study of 35 cases with a mean interval of 47 days. Meanwhile, remarkable prolongation has been reported in a few cases. The case with the longest interval between the births of both twins spanned 154 days. Aydin and Celiloglu described a case wherein, after the delivery of the first twin at 21 weeks’ gestation, a 3639g infant was delivered with cesarean section at 36 weeks’ gestation and had an uneventful neonatal course. Furthermore, some studies have reported cases of pregnancy prolongation of more than 100 days. Delay of delivery beyond 36 weeks seems relatively scarce, and our case is an additional one.

There is no established management strategy for delayed interval delivery. This technique may involve severe complications, primarily chorioamnionitis and abruptio placenta. Therefore, after the delivery of the first fetus, the patient must have clinical follow-up and blood tests for clinical complications like chorioamnionitis and placenta abruptio. Prevention of chorioamnionitis is believed to be the

**Figure 2.** Placenta of the dead twin was atrophic in comparison with placenta of the viable twin
most important key for a successful outcome. To avoid ascending infection, the cord of the first-born twin should be ligated with an absorbable suture as close to the cervix as possible, under aseptic conditions. Empiric broad spectrum antibiotic treatment should be initiated from the delivery. The control cervicovaginal cultures should be sampled on a periodic basis, and culture-directed antibiotic therapy according to the antibiogram is recommended until negative cultures are confirmed. The most controversial issue in management of delayed-interval delivery is whether cervical cerclage is necessary under these circumstances. Some authors have accentuated the positive effect of cerclage.2,6,11,14

Cerclage may provide stability to the cervix; furthermore, it may minimize the exposure of fetal membranes to vaginal bacteria and acidity. We also had a management policy of cervical cerclage, and a favorable outcome was achieved. On the other hand, some authors have taken a negative stance.2,4,10,15 No randomized controlled trials regarding the efficacy of cervical cerclage in delayed interval delivery are reported in the literature. Consequently, each situation should be individualized, depending on possible risks, patient wishes, and contraindications. In our case, we performed cervical cerclage even though the gestational age was 25 weeks, because in our country (Japan), therapeutic cerclage is usually considered a possible option if the gestational age is <26 weeks.16 Furthermore, in our case, two tocolytic agents were simultaneously administered over a prolonged period with close and careful observation for maternal complications. A Japanese study has recently suggested that maintenance tocolysis using a combination of tocolytic agents may be effective in a few cases including in women undergoing cerclage;17 however, long-term use of tocolytic agents in pregnancy can be associated with an increased risk of maternal adverse effects. Thus, in our opinion, further studies are necessary to gain a better understanding regarding the appropriate use of tocolytic agents in cases of delayed-interval delivery.

We reported a case of delayed-interval delivery, wherein prolongation of the second twin’s delivery until term with a subsequent favorable infantile outcome could be obtained. We believe that delayed-interval delivery can be effective in prolonging gestation and should be an option if the first twin developed an extreme preterm delivery.

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Delayed-interval delivery at term