

Delayed interval delivery of a second twin resulting in septic shock in the mother

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ABSTRACT

Delayed interval delivery is an option in the extremely preterm twin pregnancies, in an attempt to decrease morbidity for the remaining fetuses. We report a case of diamniotic dichorionic pregnancy complicated by premature rupture of membranes of the first fetus at 20 weeks gestation. Premature delivery of the first twin occurred nine days latter. Delayed interval delivery of the second twin was attempted using bed rest, tocolysis and antibiotics. Unfortunately, 14 days later spontaneous labour ensued and the birth took place with the mother presenting signs of chorioamnionitis. Septic shock occurred in the mother, with need of mechanical ventilation and aminergic support. It seems to us that it is important to present unsuccessful cases like this in order to discuss what are the optimal management options, while there is no universal agreement on this issue.

Keywords: delivery-obstetric; twins; septic shock

PARTO DIFERIDO DE UM GÉMEO CAUSADOR DE CHOQUE SÉTICO NA MÃE

RESUMO

O parto diferido de um gémeo é uma opção nas gestações complicadas por prematuridade extrema, como forma de tentar diminuir a morbidade dos fetos restantes. O caso aqui apresentado é de uma gravidez bicoriónica biamniótica complicada por rotura prematura de membranas do primeiro feto às 20 semanas de gestação. O parto do primeiro gémeo ocorreu nove dias depois. O parto diferido do segundo gémeo foi tentado, recorrendo a repouso, tocólise e antibióticos. Contudo, 14 dias depois, ocorreu o parto espontâneo do segundo gémeo, com a mãe a apresentar sinais e sintomas de corioamnionite. A mãe acabou por desenvolver um choque séptico, com necessidade de ventilação mecânica e suporte aminérgico. A apresentação de casos de insucesso como este parece-nos importante, porque permite discutir as opções terapêuticas, enquanto não existir um consenso universal sobre este assunto.

Palavras-chave: parto; gravidez múltipla; choque séptico

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INTRODUCTION

Multifetal pregnancies are increasing as a result of increased maternal age and increased use of medically assisted reproduction techniques. There is a high risk of preterm delivery and preterm premature rupture of membranes in multiple pregnancies, usually with the delivery of all the fetuses within a short time interval.^{1,2}

Delayed interval delivery is an option in the extremely preterm, allowing to decrease morbidity for the remaining fetuses; however, it can lead to serious maternal, fetal and neonatal morbidity and even mortality.² There is a lack of a universally accepted protocol for the management of such cases in the literature.² Most studies are case reports and small case series, the majority of which report successful outcomes.³⁻⁷ It seems that there is a bias of publication of the complications of delaying delivery. The largest series published reports 50 cases of delayed delivery and provides an overall survival of 26% in the first twin and 66% of the co-twin.²

The use of prolonged bed rest, tocolysis, antibiotics, corticosteroids and cervical cerclage is under debate, but there is no agreement regarding the best management option. We report a case of 14 days delayed interval delivery with poor maternal and fetal outcomes.

CASE PRESENTATION

Thirty six year-old healthy woman, G3P2 (two vaginal term deliveries), with spontaneous diamniotic dichorionic twin pregnancy diagnosed at 17 gestational weeks.

She was admitted to our emergency department at 20 weeks and 4 days with preterm premature rupture of membranes of the first twin. Both were cephalic and had good vitality. Cervical length was 37 mm. Her leukocyte count was $11,18 \times 10^{12}/L$, C reactive protein was 13.53 mg/dL, urine culture was negative and *Mycoplasma* spp and *Ureaplasma* spp were isolated in the cervicovaginal exsudates. Bed rest, azithromycin 1000 mg per os in single dose, amoxicillin per os 500 mg 8/8h and subcutaneous enoxaparin 40 mg once a day were prescribed.

At 21 weeks and 6 days she complained of painful uterine contractions and rapid delivery of the presenting fetus occurred, without delivery of the placenta. The fetus weighted 440 g.

Patient was informed about the possibility of attempting deferred delivery of the remaining fetus, its benefits and risks, which she accepted. The umbilical cord was ligated as high in the cervix as possible and the placenta was left inside the uterus.

Prophylactic tocolysis with nifedipine 20 mg per os 8/8h was prescribed and amoxicillin per os was maintained in the same posology. Her leukocyte count was $22,28 \times 10^{12}/L$ with neutrophilia and C reactive protein was 26.96 mg/dL. The vitality of the surviving twin was assessed by daily assessment of fetal heart sounds and ultrasound on alternate days. Seven days after the delivery of the first twin, as the patient was asymptomatic, apyretic, her leukocyte count was $13,70 \times 10^{12}/L$ and her C reactive protein was 13,00 mg/dL, we opted for suspending antibiotics and tocolysis and maintain inpatient surveillance.

At 23 weeks and 4 days, the patient complained of abdominal pain and vaginal hemorrhage: spontaneous labour had begun. Her auricular temperature was 38.9 °C. Intravenous ceftriaxone 2g and clindamycin 900 mg in single dose were initiated and we decided not to stop labour. Three hours latter delivery of the second twin occurred, a female that weighted 570 g and died soon after birth. The patient remained febrile and blood cultures were requested. Two hours after the delivery, manual removal of the placenta in the operating room was required. The patient developed dyspnea and her oxygen saturation dropped. Her blood analysis showed a leukocyte count of $42,90 \times 10^{12}/L$, a C reactive protein of 139.00 mg/dL and a lactate of 5.4 mmol/L. She was admitted in the intensive care unit with the diagnosis of septic shock, requiring invasive ventilation and norepinephrine for two days. Intravenous clindamycin 900 mg 8/8h was maintained and intravenous imipenem 1 g 8/8h was added in the next 10 days. Microbiological analysis of the placenta revealed the presence of *E. coli* and blood cultures were negative. The patient gradually improved and was discharged home 12 days later fully recovered.

DISCUSSION

Gestational age is the most important predictor of survival in infants delivered before 28 weeks gestation, which makes delayed interval delivery a reasonable option in cases such as the one we presented. The greatest potential benefit of this attempt is before 24 weeks gestation, because it allows the remaining fetuses to reach viability. On the other hand, survival rates are poorer at this gestational age because latency is often not enough to reach viability.⁸ Corticotherapy is important for fetal lung maturation, but given the gestational age, it was not initiated in this case.

Our management was similar to many others found in the literature with reported good outcomes, but the absence of unanimity about the best management of these pregnancies makes it difficult to decide which interventions to perform. There are some other steps reported in the literature that we could have done.

Some authors perform amniocentesis on the retained fetus in order to exclude subclinical amniotic infection, which precludes the attempting of delaying delivery. Any sign of intra amniotic infection, such as maternal fever, maternal or fetal tachycardia, maternal leukocytosis or uterine tenderness would have stopped this attempt. Our concern about infectious morbidity made us use antibiotics until the patient was not demonstrating any sign or symptom of infection. We could probably have made an irrigation of the lower uterine segment with antibiotics after the cord ligation and have repeated the exsudates after the delivery of the first fetus. Membrane rupture of the second twin, monochorionic placenta, *abruptio placentae*, severe pre-eclampsia or other serious complications related to pregnancy that could put the mother at risk would have prevented this management.

There are some descriptions of use of endoloop ligation of the umbilical cord under ultrasound guidance as being more effective than traditional suture with potential benefit of reducing subsequent infection risk;⁹ this needs to be further studied.

Some authors recommend use of cerclage in all delayed interval deliveries, others do not recommend it.^{2,10} In this case it was not performed, but we suppose that the pathophysiology of this preterm labor was not related to cervical incompetence but to infection.

Another important point is that these procedures should be done in tertiary centres with intensive care unit support because their performance can be lifesaving.

Our poor maternal outcome is not unique in the literature, but there are few cases described of serious maternal morbidity.¹¹⁻¹³ It seems to us that it is important to present unsuccessful cases in order to discuss what are the optimal management options, while there is no universal agreement on tocolysis, antibiotics, amniocentesis, corticosteroids, cerclage and type of umbilical cord ligation techniques.

HIGHLIGHTS

Delayed interval delivery is an option in the extremely preterm, allowing to decrease morbidity for the remaining fetuses; however, it can lead to serious maternal, fetal and neonatal morbidity and even mortality.

We report a case of 14 days delayed interval delivery with poor maternal and fetal outcomes.

Our poor maternal outcome is not unique in the literature, but there are few cases described of serious maternal morbidity.¹¹⁻¹³ It seems to us that it is important to present unsuccessful cases in order to discuss what are the optimal management options.

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