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Transfusion Syndrome in a Triplet Pregnancy

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Abstract. A case is reported of a transfusion syndrome in a triplet pregnancy with intrauterine death of two of the fetuses. This is an exceptional occurrence in a triplet pregnancy and raises the problem of the management of multiple pregnancies associated with the death of one or more of the fetuses. The problem of the method of delivery and the complications arising in triplet dizygotic pregnancies is discussed.

Key words: Triplet pregnancy, Transfusion syndrome, Intrauterine death, Zygosity, Vaginal delivery

INTRODUCTION

Triplet pregnancies are rare. Their frequency in the French population is 1/7921 according to Hellin's law [16]. There are three types of triplet pregnancies: monozygotic, dizygotic, and trizygotic.

Numerous complications specific to dizygotic triplet pregnancies are reported in the literature. The transfusion syndrome is a rare complication in monoamniotic twin pregnancies. We report a case of transfusion syndrome in a probably dizygotic triplet pregnancy. No similar case has been published previously.

CASE REPORT

A 31-year-old teacher, gravida 2, para 1, presented with a triplet pregnancy conceived naturally. Her blood group was A positive and she was seropositive for toxoplasmosis and rubella. Both patient and husband had a family history of twin pregnancies.

The patient began antenatal care at 27 weeks gestation at Antoine Béclère Hospital, in Clamart, France, at which time the triplet pregnancy was diagnosed. She had been previously followed elsewhere.

The ultrasound examination at 27 weeks revealed one membrane separating two am-

niotic sacs. Fetuses A and C were of normal sizes for the gestation period, while fetus B was found to have intrauterine gestation growth retardation. Fetuses B and C, in the second sac, were hypotrophic and of normal size, respectively. At the time of this examination neither of these two fetuses (B and C) had cardiac activity. The third fetus (A) was alive. Ultrasound examination confirmed the death of the two fetuses contained in the same sac. The amniotic fluid of the third fetus was very abundant. The patient had a normal coagulation profile. At 29 weeks and four days, the patient had premature rupture of the membranes with loss of a chocolate-coloured liquid.

An unsuccessful attempt was made to stop uterine contractions by Ritodrine perfusion. Bacteriological examination of the amniotic fluid revealed numerous erythrocytes, leukocytes and some Gram+ bacilli (Doderlein Bacilli). No antibiotics were prescribed. It was not possible to visualize at ultrasound examination which of the two sacs had ruptured.

The examination was interrupted because of painful contractions. The cervix was 8 cm dilated.

In view of the cephalic presentation of the living fetus, the risk of maternal infection, and the gestational age, it was decided to allow a vaginal delivery. The cardiac rhythm of the living fetus was normal. Full dilatation was rapidly reached.

A pudendal block was given. The first triplet was stillborn and non-macerated. The umbilical cord was engorged with blood. The amniotic sac of the second triplet was intact. Artificial rupture of the membranes was performed resulting in clear liquid.

A low forceps delivery was performed on account of inefficient maternal expulsive efforts. A male infant was delivered who cried immediately. This infant was immediately handed over to the pediatricians. The third triplet was stillborn, non-macerated, and the cord was exsanguinated.

The second triplet weighed 1200 g and had an Apgar score of 4 at 1 minute, followed by secondary apnea. He was intubated and ventilated with 100% oxygen. Hyaline membrane disease was rapidly diagnosed and treated with synthetic surfactant. This led to bronchopulmonary dysplasia and infection due to coliforms and group B streptococci. Nasogastric feeding was started on day 8. On day 12, a severe enterocolitis occurred, necessitating the discontinuation of feeding, artificial ventilation and antibiotic therapy.

On day 13, a perforation of the digestive tract occurred and 12 cm of the ileum was therefore resected. Laparotomy had to be repeated on account of two deep abcesses and necrosis of the digestive tract. The transverse colon and 40 cm of the small intestine were left after the second surgical operation. An ileostomy and a colostomy were sited.

It was envisaged to perform a reanastomosis when the child weighed 8 kg, ie, around 1 year of age. $9\frac{1}{2}$ months later the infant exhibited a normal growth and development. Neuromuscular assessment revealed the persistence of an hypertonicity of the limbs, but nevertheless there was a very clear improvement, compared with earlier examination.

Post-mortem examination of the two stillborn fetuses showed a major degree of maceration and a transfusion syndrome. The first infant was deep red, it weighed 900 g and measured 36 cm in length. The third infant weighed 800 g and measured 36 cm. It was very pale. There was no dysmorphism or visceral malformation in either of the infants.

Examination of the placentae revealed two distinct placental masses. The first placenta weighed 800 g. There was one amniotic cavity and the cord vascularised the sur-

viving triplet. Histological examination showed advanced maturity of the placental cotyledons. The second placenta was twinned, monochorionic and monoamniotic. The vascular anastomosis was superficial. Histologically, the placenta was degenerated with calcifications and a complete disappearance of villous vascularisation.

In summary, this was a case of a triplet dichorionic and diamniotic pregnancy. One of the two placentae was of the twin variety, monochorionic, monoamniotic with villous anastomosis. This abnormality is responsible for the transfusion syndrome which was the cause of intrauterine death of the two fetuses in our case.

COMMENT

The frequency of twin pregnancies is 1/89 [16]. The expected frequency of triplet pregnancies is $1/89^2$, ie, 1/7921 (Hellin's law). The incidence of triplet pregnancies at Antoine Béclère Hospital is 1/1640. The rate of triplet pregnancies compared to twins is 1/25 [15].

It is not possible to state the zygosity in our own or other clinical studies of triplet pregnancies. Derom et al [4] suggested a frequency of dizygotic pregnancies of 40%. Half were induced and the other half occurred spontaneously. The transfusion syndrome is one of the serious complications of twin pregnancies. It is also frequently implicated in the intrauterine death of one of the twins. Monochorionic monoamniotic pregnancies are specially associated with this risk. These pregnancies are rare and only represent 2% of twin pregnancies [3].

Conversely, the placental anastomosis is very rare in dichorionic-diamniotic twin pregnancies, which are more frequent (80%) [3].

In our report, the diagnosis of dizygotic triplet pregnancy is probable, but cannot be confirmed. It is not excluded that the triplets were all monozygotic, two monoamniotic and the third in a separate placenta.

The consequences of intrauterine death of one or two fetuses are serious. They can be fetal or maternal. The complications are related to problems of coagulation disorders, induced by thromboplastin originating from the fetus and placenta, the first observation of maternal complication was presented by Skelly et al [18]. It was a triplet pregnancy with intrauterine death of two fetuses and maternal disseminated intravascular coagulation (DIC). One fetus died in the second trimester. At the fifth month, maternal disseminated intravascular coagulation (DIC) occurred and was treated with heparin. The heparin corrected the DIC and the pregnancy continued. At 35 weeks, the second fetus died in utero. A cesarean section was performed at 36 weeks to extract the surviving fetus which weighed 2250 g.

This observation is one of the few to describe the consecutive death of two of the three fetuses. This case was not a dizygotic pregnancy, as three amniotic cavities were present. In twin pregnancies, the frequency of intrauterine death of a fetus is approximately 3.7/1000 [9,10]. In monozygotic twin pregnancies, the frequency is 3.7% [13].

Two risks threatened the surviving fetus [7]:

1) The persistence of the factors which cause the death of the first fetus;

2) In the case of cross circulation, the retention of the dead fetus can cause DIC in the survivor [14,18], or can cause embolic migration of necrotic placental tissue. Severe

neurological problems have been reported in the survivor which may cause neonatal death [2,6,13,22]. Cortical renal necrosis associated with cerebral and splenic infarction and thromboses of cerebral arteries have also been described [1,14].

In our report, there was no vascular communication between the surviving triplet and the two died in utero. The surviving fetus had no coagulation problems or encephalomalacia. Following the death of the two fetuses no coagulation disorders were seen in the mother.

Mode of delivery. The risks threatening the surviving fetus justify cesarean section when the pulmonary maturity appears adequate. This is specially important because the complication described can occur very quickly after fetal death [11]. Currently, most authors [2,11] suggest monitoring the pregnancy by carrying coagulation tests every 48 hours and daily assessment of the fetal heart rate. The aim is to reach fetal maturity and possible vaginal delivery.

Some authors have suggested routinely terminating these pregnancies at 36 weeks, preferably vaginally, if obstetrical facts are favourable [3,20]. Most authors advise cesarean section in triplet pregnancies [8,12,15]. This leads to a reduction in neonatal mortality and morbidity.

In our series, cesarean section was routinely performed [15]. However, in this case we decided to perform a vaginal delivery because of: 1) the presence of a single surviving fetus, 2) maternal-fetal infection, 3) the rapidity of the labour, 4) the potential risks of coagulopathy, and 5) the cephalic presentation of the surviving fetus.

The fetal heart rate was normal throughout labour.

No case of transfusion syndrome has previously been reported in a triplet dizygotic pregnancy. However, many complications of these pregnancies have been described. These complications are mainly associated with an abnormal infant and with siamese infants [5,17,19,21].

CONCLUSION

Transfusion syndrome is an exceptional complication in triplet pregnancies. Difficult problems are posed by the maternal risks and the hazards to the surviving infant. The management of the pregnancy is very delicate because of the psychological problems involved by the coexistence of one or more dead fetuses with a living one. A classical bereavement cannot occur because of the presence of a living child. The extreme anxiety on the mother's part faced with the risks for the surviving fetus make obstetrical management very difficult.

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