Maternal and neonatal outcome in a monochorionic twin pregnancy complicated by single intrauterine demise

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Summary

Single fetal death in monochorionic pregnancies is believed to be associated with increased risk of perinatal morbidity and mortality for the living twin and risk of coagulopathy affecting the mother. In this report we present a case of single intrauterine death in a monochorionic twin gestation diagnosed in the 28th week of pregnancy.

Key words: Twin pregnancy; Single fetal death; Placentation; Maternal and neonatal outcome.

Introduction

The incidence of twin pregnancies has increased remarkably since the late seventies because of the progress in reproductive medicine [1]. Unquestionably, twin pregnancies are at greater risk of adverse maternal and fetal outcome with preterm delivery and intrauterine growth retardation being the commonest factors contributing to perinatal morbidity and mortality [2, 3]. Single fetal death in the first trimester is relatively common (10%-70%) and the course of pregnancy is usually not impaired [3, 4]. By contrast, single fetal death in later pregnancy occurs only in 0.5%-7% with increased perinatal morbidity and mortality for the living twin and risk of coagulopathy affecting the mother [5, 6]. Perinatal mortality in monochorionic placentation, occurring only in about 20% of twin gestations, is about three times higher than in dichorionic twins and the prevalence of monochorionic placentation in case of a single fetal death varies between 35%-70% [6, 7, 8].

Because of the rarity of a single fetal death in twin gestation large reports are missing and general management is not well established. In this report we present the fetal and maternal outcome in a case of single fetal death in a monochorionic pregnancy occurring in the 28th week of pregnancy and we review the recent literature.

Case report

A 33-year-old woman (gravida II, para 0, 1 ectopic pregnancy) was admitted to our hospital in the 25th week of gestation because of preterm labour. By first trimester ultrasound examination a monochorionic, diamniotic twin gestation had been diagnosed. The cervix was unripe with a Bishop-score <3 and the cardiotocography (CTG) showed irregular contractions with normal fetal heart rates. The ultrasound examination revealed two appropriate for gestational age fetuses with normal amniotic fluid. During the 26th week of gestation a twin-to-twin transfusion occurred with oligohydramnios of one fetus and polyhydramnios of the second fetus with similar fetal growth. At serial CTGs fetal heart rate of both twins and doppler flow were normal and the patient did not feel any contractions.

Intrauterine fetal death of the acceptor was diagnosed sonographically in the 28th week of pregnancy after the heart beat of the second twin was not found in the CTG. The dead fetus showed polyhydramnios and a hydrops fetalis. The living donor was appropriate for gestational age and presented oligohydramnios. Arterial blood flow was normal. After intensive discussion the parents decided to continue the pregnancy. Fetal lung maturity was induced with steroids. Close surveillance of both living fetus and mother was performed by serial ultrasound and doppler examinations, fetal heart rate monitoring, and serial assessment of the coagulation status of the mother. In the further course gestational diabetes was diagnosed by a glucose stress test and a diet was followed. In the 31st week of gestation regular contractions with a cervical dilatation of 5 cm occurred. Cesarean section was performed because of breech presentation of both twins. Fetus A was an anemic female, 1560 g, 45 cm, Apgar-score 8/intubation, pH_{art}: 7.31; Fetus B was a macerated stillborn female, 1160 g, 39 cm, without gross anomalies. Histologic examination showed a monochorionic, diamniotic placenta of 630 g with normal insertion of the umbilical cord and arterio-arterial and arterio-venous anastomoses. During the neonatal period a first degree periventricular leucomalacia was diagnosed by MRI and a second degree RDS was diagnosed by X-ray. The newborn was discharged on day 61. The mother developed a thrombosis of the lower right leg in the 27th week of gestation and a hemostaseologic examination revealed an enhanced platelet aggregation on a genetic basis. At a follow-up examination after two years the infant showed normal motor development.

Discussion

Twin gestation, occurring in about one out of every 80 pregnancies, is associated with increased perinatal morbidity and mortality compared to singleton pregnancies and this difference is more important in monochorionic than in dichorionic twin gestation [7, 8, 9]. This fact is mainly due to preterm delivery and fetal growth retardation.

There is now good evidence that the early pregnancy loss in twin gestation, the vanishing twin phenomenon, occurs in about 20%-50% of all twin gestations without any adverse effect on the course of pregnancy [3, 4, 10].

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By contrast, single fetal death in twin gestation after 20 weeks of gestation is supposed to be a rare event with a supposed incidence of 2.6%-5.9% [3, 8, 11-14]. In older reports including first trimester single fetal death in twin gestation the incidence of this complication varies from 0.5%-6.8% [7, 15, 16]. In the largest single study of 29 cases Santema *et al.* found an incidence of 5.4% of single fetal death after 20 weeks of gestation [11]. In all we found 193 cases of single fetal death in twin gestation after 20 weeks of gestation published in the literature until 1998. The over-all incidence was 3.98%.

In the presented case of monochorionic, diamniotic twin gestation we suppose acute twin-to-twin transfusion to be the cause of the single fetal death. Histologic examination revealed arterio-arterial and arterio-venous anastomoses in the monochorionic placenta with the surviving donor twin being anemic at birth. Vascular anastomoses are present in almost every monochorionic placenta, but not regularly in dichorionic placentas [7, 17]. Insertio velamentosa, which is believed to be up to nine times more common with twin gestation was not observed in our case [7]. We did not find cord entanglement, which accounts for the perinatal mortality of 50% of monoamniotic twins [9].

Conflicting data exists concerning the mortality and morbidity risk of the surviving twin. The incidence of perinatal mortality of the second twin varies from 0%-46% with a higher risk for monochorionic pregnancies. [12, 18]. Enbom estimated the risk of severe morbidity or mortality of the surviving twin to be 46% [15]. Fusi *et al.* described two deaths of the second twin in eight monochorionic pregnancies (25%), Prömpeler *et al.* found in their recent report four deaths of the remaining twin (2 in monochorionic, 2 in dichorionic placentation) in a total of 32 twin pregnancies (13%) [8, 12]. In the largest single series of 29 cases, of which there were 13 monochorionic and 16 dichorionic pregnancies, in two monochorionic pregnancies the fetal co-twin died five and ten days, respectively, after the death of the first twin (7%) [11]. In our case fetal death of the second twin was not seen.

According to recent reports major neurological abnormality, e.g. cerebral infarction, multicystic encephalomalacia, was seen in 17% of the remaining twins after single fetal death in monochorionic gestation but not in dichorionic gestation [6, 9, 11]. In 1961 Benirscke supposed disseminated intravascular coagulation (DIC) in the surviving twin due to thromboplastic material from the dead fetus to be responsible for neurologic damage after fetal death in a monochorionic gestation [19]. Moore *et al.* described similar findings in three cases with renal and cerebral necrosis associated with fibrin deposits being revealed by autopsy [20].

However, DIC may not be the only cause for brain damage in the living fetus. Okamura *et al.* reported in their recent study, that cordocentesis performed in seven fetuses after the death of the co-twin revealed anemia in 5 fetuses without signs of coagulopathy [2]. The authors proposed that hypotension and anemia in the living twin caused by the fall of blood pressure in the dead twin may have been sufficient to cause neurologic injury in the surviving twin. In our case we found twin-to-twin transfusion with an anemic donor and a stillborn acceptor, which could in part be explained by hemodynamic changes with transfusion towards the acceptor twin after its death.

Although we found first degree periventricular leukomalacia in the surviving fetus with normal follow-up, we share the opinion of Santema *et al.* [11] and do not agree with Enbom *et al.* [15] that multicystic encephalomalacia is a frequent finding and only half of the surviving twins show normal development. It seems more likely to us, that intrauterine growth retardation and prematurity of the surviving twin remain the main risk factors for perinatal morbidity and mortality.

Maternal coagulopathy caused by tissue thromboplastin from the dead fetus and its placenta was first described by Weiner *et al.* in 1950 [21]. In 1955 Pritchard *et al.* reported a 25% incidence of maternal coagulation disorders in singleton pregnancies with a dead fetus retained for more than five weeks [22]. In their recent study Santema *et al.* did not find any complication caused by disseminated intravascular coagulation in four patients in whom the dead fetus remained more than five weeks in utero and they supposed the risk of maternal coagulopathy due to fetal death to be low [11]. In our case maternal coagulopathy due to the retained fetus did not occur. Thrombosis of the lower right leg occurred 10 days before the onset of single fetal death. Clotting tests revealed an enhanced platelet aggregation probably on a genetic basis. There was no maternal sepsis.

Concerning the question whether immediate delivery or expectant management after diagnosis of single fetal death in twin gestation is advantageous, debate remains controversial. It is not clear at what time and by which mechanisms the living fetus could be damaged after single fetal death and to date neurologic injury of the living twin cannot be prevented by any therapeutic intervention. Thus, aggressive management with premature caesarean section after diagnosis of the fetal death did not improve perinatal outcome [9, 14, 23].

The authors feel that expectant management before 34 weeks of gestation with close surveillance of both fetus and mother and application of steroids for fetal lung maturity seem advisible. In the presented case the delay of fetal death to delivery was 21 days. Liberal use of cesarean section in a tertiary center seems prudent for obstetrical reasons (IUGR, twin-to-twin transfusion, preeclampsia, preterm labour) which have been shown to be associated with twin gestation. Delivery should be envisaged after 37 weeks of gestation.

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