Unexpected outcome (positive or negative) including adverse drug reactions

Spontaneous monochorionic triplet pregnancy with no fetal anomaly or feto-fetal transfusion

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Summary

We present a case of spontaneously conceived monochorionic triamniotic triplet pregnancy with no fetal anomaly or feto-fetal transfusion. The mother developed gestational diabetes at 20 weeks gestation, and at 20 weeks and 4 days gestation triplet 2 was found to have selective growth restriction and absent end diastolic flow. Although laser treatment and clamping of the umbilical cord of the growth-restricted baby was considered, the management was conservative with regular ultrasound scans and antenatal corticosteroids at 24 weeks gestation, and the umbilical artery Doppler reverted to normal at 26 weeks and 4 days and growth velocity was maintained. An elective caesarean section was performed at 33 weeks gestation to deliver three boys in excellent condition who only required feeding support.

BACKGROUND

Monozygotic multiple gestations occur when a single fertilised ovum splits into genetically identical embryos. Exactly when this division occurs governs the configuration of chorion and amnion compartments, with later fission resulting in development of progressively greater shared tissue elements. Monochorionic multiple pregnancies occur when the embryo undergoes cleavage 4–7 days after fertilisation.

Triplet pregnancies account for approximately 1:7000 births and monochorionic triplets are rarer—1:100 000 births.¹ Most of the reported cases of monochorionic triplets have been conceived through assisted reproductive techniques,² and spontaneous monochorionic triamniotic triplet pregnancies are very rare indeed. In fact, to our knowledge, there are only seven other reported cases of spontaneously conceived monochorionic triamniotic triplet pregnancies, three with congenital malformation in one of the triplets and the other four developed feto-fetal transfusion with high perinatal morbidity and mortality.

This is the first reported case of spontaneous monochorionic triamniotic triplet without fetal anomaly or feto-fetal transfusion that was managed conservatively with an excellent outcome for the mother and three babies. This normal outcome may help in decision-making and counselling of parents.

CASE PRESENTATION

This 37-year-old lady from the Indian subcontinent had two children following two full-term vaginal deliveries. In her third pregnancy, she had a dating ultrasound scan at 13 weeks and 5 days gestation and was found to have a monochorionic triamniotic triplets pregnancy with an estimated date of delivery of 6 July 2012. This was a spontaneous conception, and apart from a history of varicose veins and occasional migraines, she was well. She gave a family history of diabetes, but no history of multiple pregnancies.

She was referred to and managed jointly with a tertiary fetal medicine unit. The management plan included iron and folate supplementation, biweekly ultrasound scans until 24 weeks gestation and every 4 weeks thereafter, cervical length assessment at 23 weeks gestation, antenatal corticosteroids at 28 weeks gestation and elective caesarean section at around 33–34 weeks gestation.

The antenatal course was uneventful until 20 weeks gestation, when she was diagnosed with gestational diabetes that was initially diet-controlled but required insulin from 28 weeks gestation. A detailed anomaly scan was performed at 20 weeks and 4 days, which showed selective fetal growth restriction (FGR) in triplet 2 and discordance (weight 20% lower than triplet 3 who was the biggest), absent end diastolic flow (EDF) in umbilical artery and high pulsatility index (PI) in ductus venosus. The middle cerebral artery Doppler was reported to be normal. All three babies had normal amniotic fluid volume. Cervical length was measured at 44 mm. The plan was to consider laser treatment if there was further deterioration in a week's time.

A repeat ultrasound scan a week later (21 weeks and 4 days gestation) showed 23% discrepancy in weight, intermittent absent/reversed EDF and a normal PI in ductus venosus of triplet 2. Again, all three babies had normal amniotic fluid volume. The plan was to consider clamping the umbilical cord of the growth-restricted baby if the situation got worse. At 24 weeks gestation she was administered antenatal corticosteroids under sliding scale insulin cover. Thereafter, she continued to have weekly ultrasound scans and the situation remained stable and unchanged up until 26 weeks and 4 days gestation when the ultrasound scan showed that the fetal Doppler had normalised, with positive EDF in the umbilical artery. A further scan 1 week later showed normal Dopplers. Scans done subsequently at 30 and 32 weeks gestation showed normal growth and amniotic fluid for all the babies (figure 1).

Figure 1 Graphs of estimated fetal weights.

An elective caesarean section was performed on 17 May 2012 at 33 weeks gestation. Three live male infants were delivered in good condition with Apgar scores of 9 at 1 min, 9 at 5 min and 9 at 10 min. At her request, bilateral tubal ligation (female sterilisation) was performed at the time of the caesarean section. Triplet 1 weighed 2060 g, triplet 2 weighed 1727 g and triplet 3 weighed 1890 g. All three infants were admitted to the neonatal unit for 4 weeks in view of prematurity and need for nasogastric tube feeds. At the time of discharge, all three babies were doing well on bottle feeds and were gaining weight.

OUTCOME AND FOLLOW-UP

The pregnancy was managed conservatively and an elective caesarean section was performed at 33 weeks gestation. The babies were delivered in excellent condition and were doing well at the time of discharge. They will be followed up by the neonatal team.

DISCUSSION

To our knowledge, there are seven case reports of monochorionic triamniotic triplet pregnancies following spontaneous conception. In three of these cases, one of the triplets had a congenital malformation. In the first case,¹ the diagnosis was a congenital cystic adenomatoid malformation of the lung. In the second case,³ one of the triplets was found to be acardiac. In the third case report,⁴ one of the triplets had hydropic acardia resulting in mirror syndrome. The other four cases were complicated by feto-fetal transfusion syndrome. In one case report,⁵ symptoms of a recipient (polyhydramnios, tricuspid valve insufficiency and ascites) were present in the third triplet. The second of the donor twins died in utero at 25 weeks. At 27 weeks, a caesarean section was performed mainly due to pre-eclampsia. The first donor triplet developed normally, whereas the recipient showed periventricular leucomalacia and neurological impairment. In the second case report,⁶ feto-fetal transfusion syndrome (FFTS) was noted at 18 weeks of gestation with one donor (oligohydramnios) and one receptor (polyhydramnios) triplet. The patient received a diet enriched with proteins, and amniotic-fluid volume returned to normal after 24 weeks. At 32 weeks, a caesarean section was performed because of an intrauterine growth restriction of the donor triplet. In the third case report,⁷ FFTS was noted at 21 weeks, which rapidly progressed over the next few weeks despite indomethacin and reduction amniocenteses. At 27 weeks, spontaneous labour began and three live babies were delivered by emergency caesarean section, but two of them died within 2 days of birth. In the fourth case report,⁸ one of the triplets died at 18 weeks and 6 days. The surviving triplets had ongoing FFTS and ultrasound evidence of secondary structural brain damage was noted in the donor. This was managed by bipolar umbilical cord coagulation of the donor triplet at 23 weeks, and subsequent spontaneous vaginal delivery of the recipient triplet at 40 weeks. Therefore, spontaneous monochorionic triplet is associated with high perinatal morbidity and mortality.

The current case was complicated by FGR, discordance and abnormal umbilical artery Doppler in one of the triplets, with the potential risk of demise of all the babies in

case of demise of the growth-restricted one. Hence, laser treatment or clamping the umbilical cord of the growth-restricted baby was considered. Subsequently, triplet 2's growth stabilised and his Doppler normalised. The administration of antenatal corticosteroids may have resulted in the normalisation of the umbilical artery Doppler in triplet 2. This is consistent with the findings of Barkehall-Thomas *et al*⁹ that antenatal administration of betamethasone in multiple pregnancies with umbilical artery absent end diastolic flow is associated with the return of end diastolic flow.

Learning points

- ▶ Accurate determination of chorionicity in the first trimester is important, since obstetric outcome and clinical management are dependent on this.
- ▶ Monochorionic triamniotic triplet pregnancies should be managed in conjunction with a tertiary fetal medicine unit with facilities and expertise to deal with potential antenatal complications.
- ▶ Spontaneous monochorionic triplet pregnancies are not always complicated by congenital malformation or feto-fetal transfusion.
- ▶ Conservative management with close monitoring may be beneficial and result in a good outcome for mother and babies.

Competing interests None.

Patient consent Obtained.

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