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Fetal Intra-Peritoneal Transfusion for the Management of Very Early Spontaneous Twin Anemia-Polycythemia Sequence in an Obese Patient With a Whole Anterior Placenta

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Twin anemia-polycythemia sequence (TAPS) is a rare condition in monochorionic twin pregnancies. Small intertwin placental vascular communications allow transfusion, which results in a hemoglobin difference in the twins in the absence of oligohydramnios or polyhydramnios. We report here a case of TAPS diagnosed at 17 weeks' gestation in an obese patient (BMI 42) with a whole anterior placenta. The only possible treatment at this stage of pregnancy was intra-uterine transfusion (IUT), which was repeated weekly until photocoagulation of placental anastomoses was feasible. Fetoscopic laser surgery is the only curative treatment, but is challenging in TAPS because of the absence of polyhydramnios and the presence of minuscule anastomoses. An anterior placenta and high BMI can make the procedure even more challenging. This case report demonstrates that very early and rapidly progressing TAPS with technically complicated conditions (elevated BMI and anterior placenta) can be successfully managed with IUT until laser procedure is achievable.

■ **Keywords:** intrauterine transfusion, intraperitoneal fetal transfusion, laser photocoagulation of placental anastomoses, TAPS, twin anemia-polycythemia sequence, monochorionic twin pregnancy

A 27-year-old woman, gravida 3 para 2, was referred at 17 weeks' gestation of a monochorionic diamniotic twin pregnancy for spontaneous TAPS (middle cerebral artery peak systolic velocities [MCA-PSVs] of 50 and 16 cm/sec for donor and recipient twins, respectively). There were no criteria for twin-twin transfusion syndrome (TTTS) or selective intrauterine growth restriction. Abnormal ductal flow and pericardiac effusion were observed in the anemic fetus (TAPS stage-3; Slaghekke et al., 2010). The anterior placenta showed significant differences in echogenicity between the twin vascular territories (Figure 1).

All possible management options were presented to the parents, including expectant management, pregnancy termination, selective termination, and intrauterine transfusion (either intravascular IUT-IV or intraperitoneal IUT-IP). The significant risk of fetal demise (with and without procedure) was also acknowledged. After extensive counseling, the parents opted for antenatal treatment.

Due to maternal obesity (BMI 42, 15 cm maternal-ombilic-to-amniotic cavity distance) and placenta completely covering the anterior uterine cavity, laser surgery was not possible at this stage of pregnancy. Due to poor visibility of the thin umbilical cord or intrahepatic portion of the umbilical vein, the anemic fetus underwent an IUT-IP using 20 ml (Bowman, 1978) of packed red blood cells (hematocrit 86%) at 18 weeks' gestation. MCA-PSVs, as well as the ductal flow of the anemic twin, returned to normal 48 hours after the transfusion, while MCA-PSVs of the

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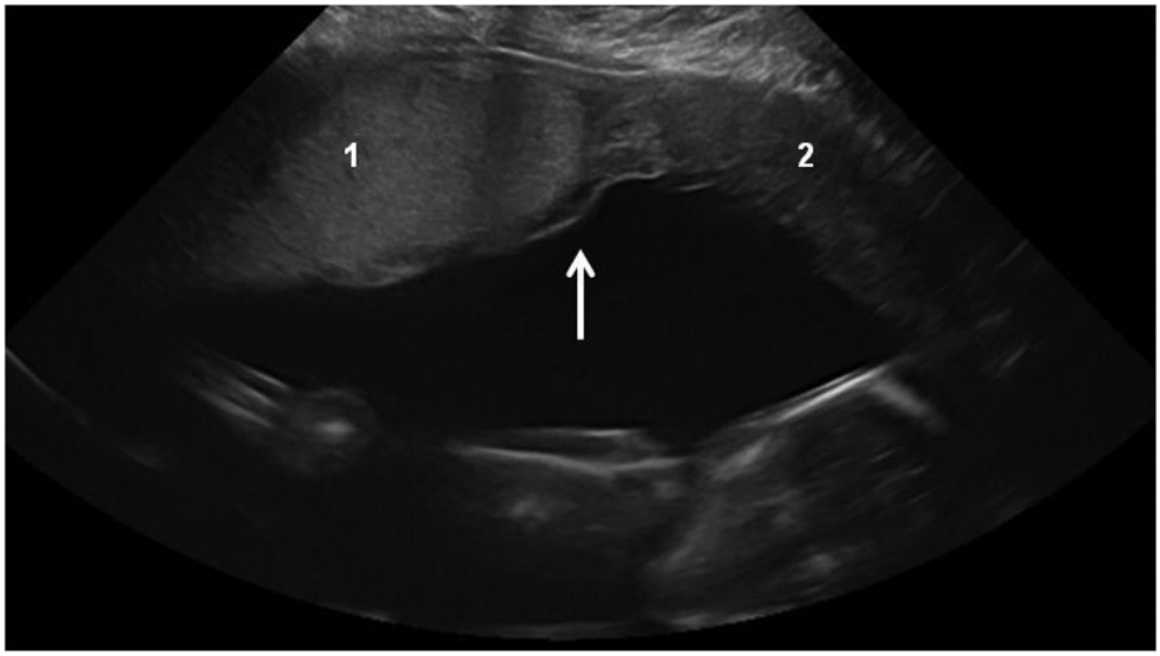

FIGURE 1

Ultrasound images at 18 weeks' gestations. Poor visibility induced by maternal obesity and a complete anterior placenta. The complete anterior placenta showed significant differences in echogenicity between the twin vascular territories, with a thick echogenic placenta for the anemic fetus and thin hypoechogenic for the polycythemic fetus. Of note, fetal karyotype on amniotic fluid was 46,XY. An arrow shows the separation between the placenta of the anemic and the polycythemic foetuses. 1: Placenta part of the anemic fetus. 2: Placenta part of the polycythemic fetus. An arrow shows the separation between the placenta of the anemic and the polycythemic fetuses.

co-twin showed only minimal changes. Ultrasound surveillance was subsequently performed every 48–72 hours.

Two additional IUT-IPs (25+30 ml at respectively 19 and 20 weeks' gestation) driven by worsening of donor MCA-PSVs were performed at 1-week intervals (Figure 2). At 21.6 weeks, null A-wave of ductus venosus blood flows, pericardic and pleural effusions were observed in both twins. Photocoagulation of placental anastomoses and all options mentioned above were discussed again with the couple, resulting in the decision to proceed with photocoagulation.

An amnioinfusion of 3 liters allowed a left lateral abdominal access for trocar insertion next to the anterior placenta. Selective laser photocoagulation of 15 fetal anastomoses was performed, completed with the Solomon technique.

A diffusion-weighted sequence magnetic resonance imaging (DWI-MR), used to detect severe brain lesions 24–96 hours after fetoscopic laser coagulation, was normal (Bebbington, 2014; Weisz et al., 2014). Serial ultrasounds showed resolution of pericardiac and pleural effusions, normal MCA-PSVs and fetal growths. Prophylactic lung maturation was done at 28 weeks' gestation. The patient was hospitalized at 29 weeks' gestation secondary to PPROM. TTTS and fetoscopic laser coagulation being associated with neurological morbidity (Huisman et al., 2005), neurosonogram and fetal MRI were planned at 32 weeks to exclude late central nervous system lesions. However, an emergency cesarean delivery was performed due to onset of labor, bleeding and breech presentation at 31+3 weeks' gestation. Placental examination confirmed chorioamnionitis and the absence of residual anastomoses after color dye injection. Birth weights (1490 g/1480 g, 35th percentile), APGAR scores, cord gases, and hemoglobin values (146/155G/l) were all normal for the ex-anemic/polycythemic twins, respectively. Neonatal brain ultrasounds were normal, neonatal follow-ups were uneventful and, at the time of writing, both children are doing well at 12 months of age.

Discussion

TAPS is a rare complication of monochorionic twin pregnancies, characterized by large intertwin hemoglobin differences due to blood transfusion through minuscule arteriovenous placental anastomoses (Slaghekke et al., 2010). TAPS may occur spontaneously or after incomplete coagulation of vascular anastomoses in cases of TTTS, with incidences up to 5% and 16%, respectively (Slaghekke et al., 2010). The antenatal diagnosis of TAPS is based on the discordance of the MCA-PSVs (>1.5 MoM and <0.8 MoM), whereas postnatal diagnosis is based on intertwin hemoglobin difference (>8.0 g/dl) and reticulocyte count ratio (>1.7) or only <1mm placental vascular anastomoses (Robyr et al., 2006; Slaghekke et al., 2010). Our case met the antenatal criteria of TAPS, with an early onset at 17 weeks' gestation, and was classified TAPS stage 3 at trans-

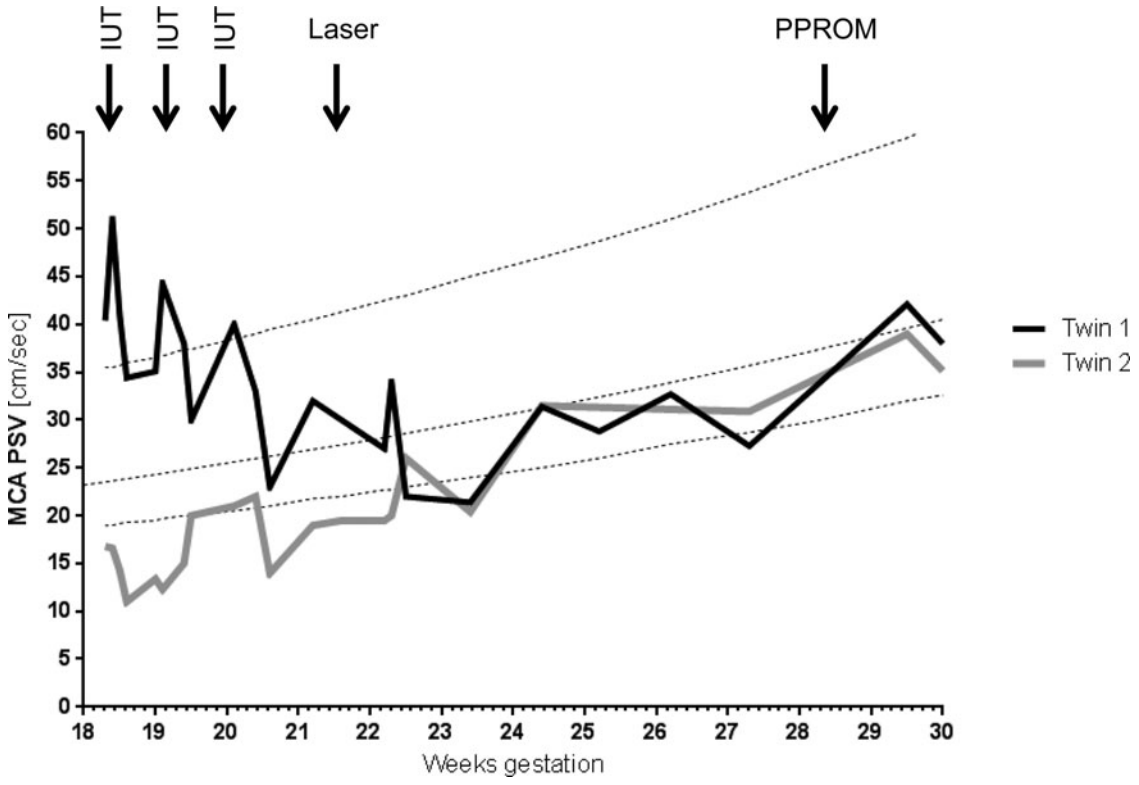

FIGURE 2

Middle cerebral artery peak systolic velocities (MCA-PSVs) measured by Doppler studies. Anemic donor in black. Polycythemic recipient in grey. A 0.8, 1.0 and 1.5 MoM grey lines are also shown. IUT-PT: intrauterine intraperitoneal transfusion.

fer 1 week later according to the classification of Slaghekke et al. (2010). Early TAPS (<18 weeks' gestation) might be suspected in case of MCA-PSV discrepancies.

Antenatal management of this condition remains controversial (Genova et al., 2013; Herway et al., 2009; Sananes et al., 2015; Slaghekke et al., 2014; 2015): expectant management, photocoagulation of placental anastomoses, IUT-IV (with or without partial exchange transfusion in the recipient; Genova et al., 2013; Slaghekke et al., 2015), IUT-IP, and elective delivery when vitality is reached. Expectant management could lead to double intrauterine fetal demise or severe cerebral injury (Lopriore et al., 2013). Photocoagulation of placental anastomoses is the only therapeutic treatment for TAPS, but fetoscopy can be technically challenging due to: (1) the absence of polyhydramnios and stuck twin as in TTTS, (2) the poor visualization of the equator due to the floating membrane, and (3) the presence of only minuscule anastomoses (Sananes et al., 2015). An amnioinfusion, as described in our case, might significantly help the fetoscopy. Whereas IUT is not therapeutic, it can be a transient alternative when fetoscopic laser treatment is not an option. However, IUT at early gestational ages of pregnancy means repetitive transfusions to reach viability.

IUT can be combined with partial exchange transfusion (Genova et al., 2013; Slaghekke et al., 2015) to reduce the severity of polycythemia in the recipient, and reduce the risk of limb necrosis and severe cerebral injury (hyperviscosity syndrome; Robyr et al., 2006). Vascular access may be technically difficult in the early second trimester of pregnancy and, when successful, may exacerbate polycythemia into the recipient twin (Lopriore et al., 2013). Robyr et al. (2006) described two fetal deaths among nine cases of TAPS after IUT-IV. IUT-IP can be performed as early as 15 weeks' gestation, as described for the treatment of early onset rhesus immunization (Howe & Michailidis, 2007). IUT-IP allows a slower absorption of red blood cells into fetal circulation (8–12 days), preventing rapid loss of the transfused blood in the circulation of the recipient twin (Slaghekke et al., 2010).

IUT-IP has already been successfully used in TAPS (Herway et al., 2009). Herway et al. described a TTTS case treated with laser surgery at 18 weeks' gestation, which developed TAPS 2 weeks later. Two IUT-IPs prolonged the pregnancy past viability, with ultimate delivery at 32.4 weeks.

Early complications of monochorionic twin pregnancies (Baud et al., 2013), anterior placenta and obese patients are all challenging situations for performing fetoscopic procedures. We demonstrate here that IUT-IP might be considered as a treatment option when fetoscopic laser treatment is not feasible, especially in cases of very early onset of TAPS, placenta covering the whole anterior uterine wall, and obese patients. IUT-IP was preferred to IUT-IV both for technical reasons and its slow absorption of red blood cells in the

fetal circulation, thus decreasing the rapid transfer of blood in the plethoric fetus. Serial IUT-IPs allowed delaying almost 3 weeks until successful laser treatment was feasible. Moreover, the longitudinal MCA Doppler studies did not show a decrease of the 'recipient' twin PSVs after IUT-IP of the anemic twin. We do not know the reason for the early hydropic signs seen at 22 weeks. Hydrops might be secondary to fluid overload (mainly for the recipient twin) and/or severe anemia (for the donor twin) despite normal MCA-PSVs 1 week after the third IUT-IP. Indeed, MCA-PSV is not useful in predicting severe anemia in fetuses that already had two previous transfusions due to varying proportions of adult blood cells in the fetal circulation that might alter fetal blood viscosity (Scheier et al., 2006).

A number of complications have been described with IUT-IP in the 1980s (Howe & Michailidis, 2007), including abdominal hematoma, and infusion within the fetal bowel or in the retroperitoneal space, but these complications have been extremely rarely reported in the recent literature (Canlorbe et al., 2011). Since IUT-IP is not a curative procedure, the recipient polycythemic twin is at risk of cardiac overload, hyperviscosity, limb necrosis, and cerebral injury due to the persistence of the TAPS.

In conclusion, IUT-PT is a viable therapeutic option in TAPS or, as shown in our case report, an opportunity to prolong pregnancy either, to reach viability or – in difficult cases – propose laser surgery.

Patient Approval

The patient whose case is described here consented to publication.

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