Clinical Case Report

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A new method for selective reduction in TRAP sequence with a contraindication to fetoscopic surgery

A case report

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Abstract

Rationale: The preferred method for multifetal pregnancy reduction (MFPR) is a transabdominal intrathoracic or intracranial injection of potassium chloride (KCl). However, in monochorionic multiple pregnancies (MMPs), especially in monoamnionic multifetal pregnancies, selective feticide by this method is often associated with miscarriage of the remaining fetuses. Selective fetal reduction in MMPs by blood flow ablation using radiofrequency ablation or fetoscopic laser surgery may improve survival of the remaining fetus. Although often successful, MFPR by these methods is contraindicated in cases of twin reversed arterial perfusion (TRAP) sequence in triplet pregnancies complicated by polyhydramnios or anterior placenta, as it is difficult to locate the ablation target.

Patient concerns: 2 cases were admitted to Xiangya Hospital, Central South University with triplet pregnancies at 23 or 21weeks of gestation.

Diagnoses: Case 1 was a 29-year-old woman with a triplet pregnancy in 2 distinct amniotic sacs and 1 fetus with multiple malformations. Case 2 was a 32-year-old woman who was identified as a triplet pregnancy with TRAP sequence with an acardiac/acephalic twin and anterior placenta.

Interventions: Both of the 2 cases were underwent a new method for MFPR involving fine needle amniotic fluid aspiration and injection of hypertonic sodium chloride (10% NaCl) into the Wharton jelly of the umbilical cord.

Outcomes: The 2 cases resulted in selective feticide and the birth of the remaining infants from the triplet pregnancies. All infants were healthy at birth and the 2-year follow-up.

Lessons: The new approach provided a safer, more accessible, and more cost-effective method for MFPR in MMPs with a contraindication to fetoscopic surgery compared to radiofrequency ablation and fetoscopic laser surgery.

Abbreviations: KCl = potassium chloride, MFPR = multifetal pregnancy reduction, MMP = monochorionic multiple pregnancy, NaCl = sodium chloride, TRAP = twin reversed arterial perfusion, USG = ultrasonography.

Keywords: case report, fetal reduction, hypertonic sodium chloride, twin reversed arterial perfusion sequence

1. Introduction

Multifetal pregnancy reduction (MFPR) is an effective measure for reducing the substantial maternal and neonatal morbidity and mortality associated with multifetal pregnancies.[1,2] Monochorionic multiple pregnancies (MMPs) present unique challenges for selective fetal reduction because of vascular anastomoses between fetuses. In these cases, MFPR can be successfully achieved by interrupting the blood flow of the communicating vessels using radiofrequency ablation or fetoscopic laser surgery.[3,4] However, in cases of twin reversed arterial perfusion (TRAP) sequence in triplets in pregnancies complicated by polyhydramnios and anterior placenta, the ablation target can be difficult to locate. We report a new method for the management of TRAP sequence in cases with a contraindication to fetoscopic surgery that involves selective reduction of the acardiac/acephalic fetus by fine needle amniotic fluid aspiration and injection of 10% sodium chloride (NaCl) into the Wharton jelly. To the best of our knowledge, this is the 1st published report to describe a method for MFPR in TRAP sequence that avoids subsequent miscarriage of the surviving fetus.

2. Case reports

2.1. Case 1

A 29-year-old woman, gravid 1 para 0 (G1P0), had undergone in vitro fertilization and embryo transfer due to primary infertility. Two fresh embryos were transferred on September 6, 2010. At 16 and 18 weeks of gestation, ultrasonography (USG) showed a twin pregnancy. At 23 weeks of gestation, the woman

Editor: Shizhang Ling.

The authors have no funding and conflicts of interest to disclose.

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Medicine (2017) 96:10(e6250)

Received: 9 August 2016 / Received in final form: 6 February 2017 / Accepted: 7 February 2017

http://dx.doi.org/10.1097/MD.0000000000006250

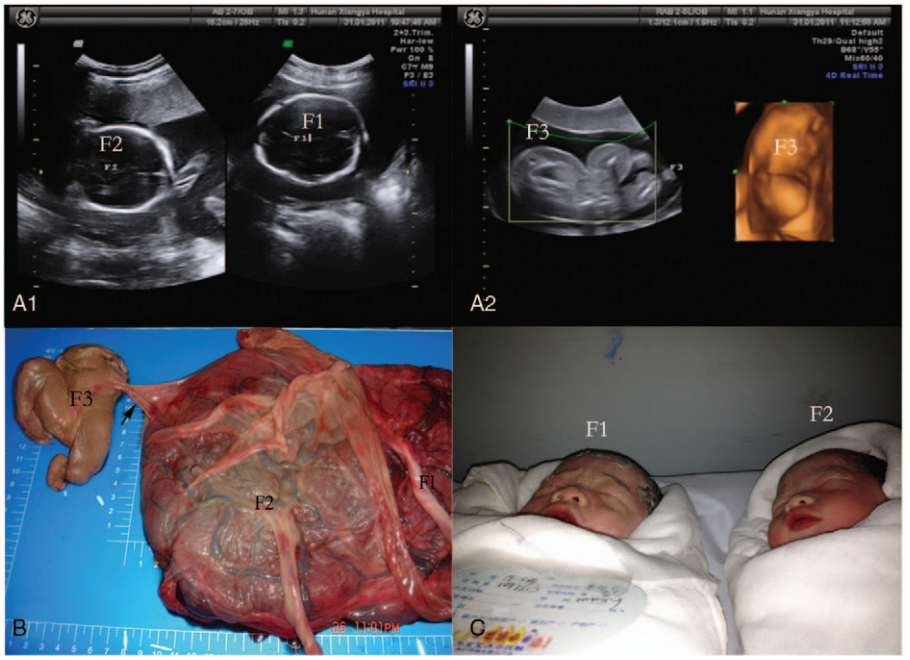

Figure 1. Case 1: ultrasound images of the fetuses before reduction and of placental vascular anastomoses, and photographs of the newborns. (A1–A2) The triplets: A1, 2 normal fetuses (F1 and F2); A2, a headless fetus with no upper and lower limbs (F3), bending deformity and polyhydramnios. (B) Placental vascular anastomoses (arrow) between the malformed and normal fetus (F2, the donor fetus) in the monochorionic placenta of the triplet pregnancy after preterm delivery. F1 was located in a distinct amniotic sac with no vascular anastamoses with F2 or the malformed fetus. (C) Normal 2 newborns.

experienced abdominal distention and regular contractions. USG revealed a triplet pregnancy with 2 distinct amniotic sacs. Two fetuses (F1, F2) had a normal fetal heart rate and amniotic fluid (Fig. 1A1). F3 was malformed with dysplasia of the spine and lower limbs; complete absence of the head, heart, and upper limbs; and hydrops (Fig. 1A2). The amniotic fluid index was 28 cm. Vascular anastomoses between F3 (the recipient) and F2 (the donor) were confirmed by color Doppler. TRAP sequence complicated with polyhydramnios was identified. Intrathoracic selective reduction of F3 was impossible due to the fetal anomaly, and cord occlusion using a fetoscope was contraindicated due to polyhydramnios and uterine contractions.[5] Therefore, after obtaining consent from the patient, we performed transabdominal fine needle (PAN Biopsy needle, 22 G × 15 cm; Gallini Medical Devices, Italy) puncture to extract 1500 mL amniotic fluid from F3s gestational sac and reduce abdominal pressure. Under color Doppler guidance (GE E8, the United States of America), we injected 15 mL of 10% NaCl into the Wharton jelly within F3s umbilical cord, which was located in the fetal abdominal wall. Three days later, USG detected no arterial flow in F3. Transabdominal cord blood puncture of F2 and F3 revealed a normal karyotype and routine hematology. At 35 weeks of gestation, the patient delivered 2 live female infants weighing 2100 and 2400 g (Fig. 1C). Placental examination revealed that F3s blood supply was provided by F2, originating from a main vessel and 2 collaterals (Fig. 1B). Apgar scores of the 1st born infant were 9 at 1 minute and 10 at 5 and 10 minutes; Apgar scores of the 2nd born infant were 7 at 1 minute and 10 at 5 and 10 minutes. Both infants were admitted to the neonatology department because of low birth weight, but they were not diagnosed with acute respiratory distress syndrome and were not admitted to the neonatal intensive care unit. The infants were discharged from the hospital after 7 days. At the 2-year follow-up, the children were healthy. The growth indices, weight, height, head circumference, and motor and language development of both children were normal.

2.2. Case 2

A 32-year-old woman, gravid 2 para 0 (G2P0), was transferred to our hospital at 21 weeks of gestation. The woman had a history of abortion at 24 weeks of gestation following a natural pregnancy. Subsequently, she underwent in vitro fertilization and embryo transfer due to secondary infertility. On March 9, 2011, 2 fresh embryos were transferred, and at 12 weeks of gestation, USG showed a twin pregnancy. At 21 weeks of gestation, USG showed a triplet pregnancy with 2 distinct amniotic sacs and placenta previa, in which the placenta was widely attached to the anterior wall of the uterus and extended to the posterior uterine wall. TRAP sequence with an acardiac/acephalic twin was identified. After obtaining consent from the patient, intrathoracic selective reduction of the acardiac/acephalic twin was performed by injecting 15 mL of 10% NaCl into the Wharton jelly within the fetus's umbilical cord, which was located in the fetal abdominal wall. Three days later, USG detected no arterial flow in the fetus's umbilical cord. Transabdominal cord blood puncture of the acardiac/acephalic fetus revealed a normal karyotype. Despite regular antenatal care, USG revealed persistent central placenta previa. At 35.6 weeks of gestation, the patient underwent an emergency cesarean section due to vaginal bleeding caused by placenta previa. She delivered 2 live male infants weighing 2050 and 1900 g with Apgar scores of 10 at 1 and 5 minutes. The infants were not diagnosed with acute respiratory distress syndrome and were not admitted to the neonatal intensive care unit. One infant had mild anemia at birth. The other infant underwent dilatation of the lower segment of the left ureter due to moderate ureterostenosis at 6 months of age. At

the 2-year follow-up, the growth indices of both children were normal.

3. Discussion

It is widely recognized that assisted reproductive technology is associated with multiple pregnancies.[2] The fetuses in multiple gestations are vulnerable to various fetal and maternal complications. MFPR, which was first introduced by Dumes and Oury for pregnancy reduction between 8 and 11 weeks, is an effective measure to reduce the complications associated with multifetal pregnancies.[1] Traditionally, MFPR involved minisuction curettage through the cervical canal under USG guidance. Technically, this is easier to perform before 9 weeks of gestation, but pregnancy loss following the technique occurs in approximately 5% of cases.[1] Presently, the preferred method for MFPR is a transabdominal intrathoracic or intracranial injection of potassium chloride (KCl).[6,7] Evidence suggests these methods are effective at 14 to 23 weeks of gestation for multifetal pregnancies derived from multiple eggs.[8] However, pregnancy loss rates of 7% to 9% for triplets and quadruplets and 42.9% for 6 or more fetuses following intrathoracic injection of KCl have been reported.[7] In addition, the donor fetus (pump fetus) in TRAP sequence may be affected by the fetotoxicity of KCl. Therefore, based on the knowledge that intravenous injection of 3% NaCl can reduce cerebral edema in children,[9,10] we injected a higher concentration of 10% NaCl into the Wharton jelly for the selective reduction of the abnormal fetus in TRAP sequence. We assumed that 10% NaCl would increase the osmotic pressure of the Wharton jelly, which would absorb water from the umbilical vessels. This would elevate the pressure of the Wharton jelly and block the flow of the umbilical artery of the abnormal fetus.

In MMPs, currently available MFPR procedures are associated with miscarriage of all fetuses. Recent technical advances, such as radiofrequency ablation and fetoscopic laser surgery (laser coagulation and cord occlusion), have allowed successful selective fetal reduction when vascular anastomoses connect fetal circulations.[3,4,8,11] However, these procedures require specialized equipment and technical expertise and may not be suitable in all cases. Furthermore, fetoscopy-guided umbilical cord ligation or laser occlusion can induce a high rate of premature rupture of membranes, preterm delivery, intraamniotic infection, and bleeding, and fetoscopy is associated with bleeding in 18% of cases at a median time of 9 weeks after MFPR procedures.[11]

These considerations were particularly relevant in our study, which included 2 cases of TRAP sequence with contraindications to fetoscopic surgery. The women were admitted to our hospital with triplet pregnancies at 23 and 21weeks of gestation, respectively. In each case, 2 fetuses were normal, and 1 was acephalic and acardiac. In the 1st case, the pregnancy was complicated by polyhydramnios and uterine contractions; in the 2nd case, the pregnancy was complicated by a placenta that was widely attached to the anterior wall of the uterus. Vascular anastomoses between the donor fetus and the malformed fetus were present. In this situation, the preferred therapy, according to the previously described methods, is to block the blood supply to the malformed fetus from the donor fetus via umbilical cord ligation, occlusion by fetoscopic surgery, or radiofrequency ablation.[12] However, these methods were contraindicated due to the clinical characteristics (polyhydramnios, uterine contractions, and anterior placenta) of the 2 cases. Therefore, we injected 15 mL of 10% NaCl into the Wharton jelly of the umbilical cord of the malformed fetus. Three days after the procedure, color Doppler revealed that blood flow from the pump fetus was blocked. USG showed that growth of the malformed fetus had ceased. We propose that injection of 10% NaCl into the Wharton jelly results in high pressure that reduces or blocks the blood supply. Ultimately, as a result of our MFPR procedures, 2 families obtained healthy newborns weighing >1500 g at >35 weeks of gestation. Newborns were delivered at 13.6 gestational weeks following fetal reduction. These gestational periods are longer than the average times to delivery following radiofrequency ablation and fetoscopic laser surgery.[13,14] Therefore, fine needle amniotic fluid aspiration and injection of 10% NaCl can be considered an option in TRAP sequence when other MFPR methods are contraindicated.

In conclusion, injection of 10% NaCl into the Wharton jelly can be performed for the management of TRAP sequence in cases with a contraindication to fetoscopic surgery. It is technically easier and cheaper than fetoscopic laser surgery or other ablation methods. Furthermore, it can offer a better chance of survival of the remaining fetuses in cases of twins with vascular anastomoses compared to intracranial or intracardial KCl injection. To the best of our knowledge, the 2 cases described here are the first reported cases of fine needle amniotic fluid aspiration and injection of hypertonic NaCl (10% NaCl) into the Wharton jelly for MFPR in MMPs. Notably, this report includes 2 cases only; further studies are required to provide clinical verification of this procedure.

References

[1] Dumez Y, Oury JF. Method for first trimester selective abortion in multiple pregnancy. Contrib Gynecol Obstet 1986;15:50–3.
[2] Nanthakomon T, Chanthasenanont A, Somprasit C, et al. Twin reversed arterial perfusion (TRAP) sequence: a case report and review of treatment. J Med Assoc Thai 2015;98(Suppl 3):S132–40.
[3] Bebbington MW, Danzer E, Moldenhauer J, et al. Radiofrequency ablation vs bipolar umbilical cord coagulation in the management of complicated monochorionic pregnancies. Ultrasound Obstet Gynecol 2012;40:319–24.
[4] Takano M, Murata S, Fujiwara M, et al. Experience of fetoscopic laser photocoagulation and cord transection for twin-reversed arterial perfusion sequence. J Obstet Gynaecol Res 2015;41:1326–9.
[5] Rossi AC, D'Addario V. Umbilical cord occlusion for selective feticide in complicated monochorionic twins: a systematic review of literature. Am J Obstet Gynecol 2009;200:123–9.
[6] Lembet A, Selam B, Bodur H, et al. Intracranial injection with KCl: an alternative method in selected cases of multifetal pregnancy reduction. Fetal Diagn Ther 2009;26:134–6.
[7] Li R, Chen X, Yang S, et al. Retain singleton or twins? Multifetal pregnancy reduction strategies in triplet pregnancies with monochorionic twins. Eur J Obstet Gynecol Reprod Biol 2013;167:146–8.
[8] Lipitz S, Shulman A, Achiron R, et al. A comparative study of multifetal pregnancy reduction from triplets to twins in the first versus early second trimesters after detailed fetal screening. Ultrasound Obstet Gynecol 2001;18:35–8.
[9] Yildizdas D, Altunbasak S, Celik U, et al. Hypertonic saline treatment in children with cerebral edema. Indian Pediatr 2006;43:771–9.
[10] Kochanek PM, Carney N, Adelson PD, et al. Guidelines for the acute medical management of severe traumatic brain injury in infants, children, and adolescents – second edition. Pediatr Crit Care Med 2012;13:S1–82.
[11] Roman A, Papanna R, Johnson A, et al. Selective reduction in complicated monochorionic pregnancies: radiofrequency ablation vs. bipolar cord coagulation. Ultrasound Obstet Gynecol 2010;36:37–41.
[12] Bharadwaj MK, Mohana Priya N. A rare complication of monochorionic twin pregnancy: twin-reversed arterial perfusion (TRAP) sequence. Med J Armed Forces India 2015;71(Suppl 1):S114–5.
[13] Skiadas CC, Missmer SA, Benson CB, et al. Impact of selective reduction of the monochorionic pair in in vitro fertilization triplet pregnancies on gestational length. Fertil Steril 2010;94:2930–1.
[14] Vela G, Luna M, Barritt J, et al. Monozygotic pregnancies conceived by in vitro fertilization: understanding their prognosis. Fertil Steril 2011;95: e606–10.