

Bipolar Cord Coagulation in Monochorionic Twins Discordant for Major Fetal Anomalies

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Abstract

Objective: We have aimed to evaluate perinatal outcome of selective fetocide in monochorionic diamniotic twins discordant for major fetal anomalies.

Materials and Methods: The study group included nine cases of monochorionic diamniotic twin (triplet) pregnancies discordant for major fetal anomalies including anencephaly (n=2), inencephaly (n=1), lumbar meningocele (n=1), hydrops fetalis (n=1), acardiac twin (n=2), twin-to-twin transfusion syndrome (stage III, n=1) and monochorionic triamniotic triplet discordant for anencephaly (n=1). For bipolar coagulation of the umbilical cord, we used 3.5 mm laparoscopic trocar and 3.0 mm bipolar forcep. The procedures were performed under the guidance of transabdominal ultrasonography.

Results: The mean gestational age of the cases at intervention and at delivery were 20 weeks (R=16-24) and 34 weeks (R=22-38), respectively. The procedure of bipolar coagulation was carried out in mean 20 min (R=15-40 min) without maternal complication. One case was aborted at 20 weeks of gestation because of preterm premature rupture of membrane after the procedure. Another case was delivered before 32 weeks, at 28+4 week of gestation, and died six months postnatally which was unrelated to the intervention. The other seven cases were delivered after 32 gestational weeks. In eight cases, the procedures were performed without fetal complication. Perinatal outcome was as 88.8% (n=8/9) live births. Postnatal ages of surviving seven babies were between 8-40 months, and they have been growing up as healthy.

Discussion: Ultrasonographic guided bipolar cord coagulation as a selective fetocide can be successfully performed to increase the survival rate or prevent morbidity of healthy co-twin in the cases of monochorionic diamniotic (triplets), discordant major fetal anomalies candidate for selective termination, and should be offered as a choice of prenatal therapy after the parents were informed of the intervention or follow up.

Keywords: monochorionic diamniotic twin pregnancy, bipolar cord coagulation, selective fetocide

Özet

İkiz Eşinde Majör Fetal Anomali Olan Monokoryonik Gebeliklerde

Bipolar Kord Koagülasyonu

Amaç: Monokoryonik diamniyotik ikiz gebeliklerde ikiz eşinde majör fetal anomali olan fetüslerde bipolar kord koagülasyonu ile selektif fetosid sonuçlarını değerlendirmek.

Materyal ve Metot: Perinatoloji konseyinde tartışılan ve tıbbi terminasyon kararı verilen monokoryonik diamniyotik ikiz gebeliklerde selektif fetosid için 3.5 mm fetal endoskopik trokar ve 3.0 mm bipolar forseps ile ultrasonografi eşliğinde bipolar kord koagülasyonu.

Sonuçlar: İkiz eşi majör fetal anomalili dokuz monokoryonik diamniyotik ikiz gebeliğe ultrasonografi eşliğinde bipolar kord koagülasyonu uygulandı. İkiz eşinde fetal anomalisi olan anensefali (n=2), inensefali (n=1), lumbar meningomyelosel (n=1), hidrops fetalis (n=1), akardiyak ikiz (n=2), ikizden ikize transfüzyon sendromu (evre III, n=1), monokoryonik triamniyotik üçüz eşi anensefali (n=1) olguları çalışma grubunu oluşturdu. Bipolar kord koagülasyonu ortalama 20 (R=15-40) dakikada gerçekleştirildi. İşlem sırasında maternal komplikasyon gözlenmedi. Olguların işlem sırasındaki ve doğumdaki ortalama gebelik haftaları sırasıyla 20 hafta (R=16-24) ve 34 hafta (R=22-38) idi. Bir olguda işlem sonrası membran rüptürü ve sağlıklı

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fetüsün eksitus olması üzerine 20 haftada abort yaptırıldı. Bir olguda doğum 32 haftadan (28+4) önce gerçekleştirildi ve bu bebek postnatal altıncı ayında kaybedildi. Yedi olguda doğum 32 hafta sonrasında gerçekleştirildi. İkiz eşi majör fetal anomalili dokuz olgudan sekizinde bipolar kord koagülasyonu maternal ve fetal komplikasyon olmadan başarı ile tamamlanmıştır. Perinatal canlı doğum oranı %88.8 olarak gerçekleşmiştir. Bebeklerin (n=7) yaşları 8-40 ay olup, şu ana kadar sağlıklı büyümeye ve gelişmeye devam etmektedirler.

Tartışma: Monokoryonik diamniyotik ikiz (üçüz) eşi majör fetal anomalili gebeliklerde, monokoryonik diamniyotik ikiz eşine tıbbi terminasyon ya da sağlıklı fetüsün yaşam şansını artırmaya ve morbiditeyi azaltmaya yönelik selektif fetosid için ultrasonografi eşliğinde bipolar kord koagülasyonu başarı ile uygulanabilen bir yöntemdir ve muhtemel komplikasyonları tartışılarak ailelere prenatal tedavi seçeneği olarak sunulmalıdır.

Anahtar sözcükler: monokoryonik diamniyotik ikiz gebelik, bipolar kord koagülasyonu, selektif fetosid

Introduction

Monochorionic twins almost always have vascular communications between the two fetal circulations, generally across the placenta. Monochorionic twinning may be complicated by a number of rare conditions, including twin reversed arterial perfusion sequence and twin to twin transfusion syndrome. Additionally monochorionic twin pairs may be discordant for structural or genetic anomalies, for which they are at higher risk than singletons (1). In all these conditions selective fetocide may be contemplated for selected cases.

The standard technique of selective fetocide in dichorionic twins of intra-cardiac KCl injection cannot be used in monochorionic twins due to high risk of the co-twin exsanguinating into the terminated twin leading to death or neurological sequelae seen in approximately 50% of cases (2). Instead, isolation of the terminated twin's vasculature is necessary by an occlusive method. A variety of ultrasound-guided and fetoscopic techniques have been used with mixed results (3). Injection of sclerosants like absolute alcohol or enbucilate gel, is now considered contraindicative because of high failure rates. The same applies to other techniques such as thrombogenic coils that occlude only one cord vessel.

Therefore, a number of minimally invasive procedures were developed to produce complete circulatory confinement of the affected fetus. Fetoscopic laser cord coagulation has been performed as early as 16 weeks. At later gestational ages, after 20 weeks, however, it may be difficult to coagulate the umbilical cord because of increasing size of the cord vessels (4). After 20 weeks, ultrasound guided bipolar cord coagulation is the preferred method. This article describes our experience with the use of a 3.0 mm forceps for a variety of indications that involved complicated monochorionic twin pregnancies.

Materials and Methods

This is a prospective follow-up study of monochorionic diamniotic pregnancies that underwent bipolar cord coagulation as a primary procedure in our prenatal diagnosis

and therapy unit. All cases were discussed with the perinatal-neonatal and hospital ethic committee before the procedures, and all families were informed on the intervention and the follow up with individual complications and outcomes. For ligation of the umbilical cord, we used 3.5 mm laparoscopic trocar and 3.0 mm bipolar forceps (Karl Storz, Tuttlingen, Germany). The procedure was carried out in the operating room, after achievement of complete aseptic conditions and 50 mg meperidine im for sedation and local anesthesia by injection of 1% lidocaine deep into myometrium. The port insertion site was chosen according to the position of the fetus and location of the placenta, and the target umbilical cord. The trocar was inserted into amniotic cavity under ultrasonographic guidance. Bipolar forceps was passed through the trocar and the portion of the umbilical cord closest to the placental insertion site was grasped. Direct contact with the placenta, fetus or membranes was avoided. Bipolar coagulation was started at 20 W and applied for 30 seconds with progressive increments of 5 W until the appearance of steam bubbles indicative of local heat production and hence tissue coagulation between the forceps' blades (usually between 25-50 W). Confirmation of arrest of flow was performed by color Doppler distal to the occlusion after the forceps was freed from the cord. Two additional cord segments were coagulated [preferentially more proximal towards the target fetus] as a safety precaution, even if there was no longer any visible flow.

Routine prophylactic measures included indomethacin tocolysis for 48 hours and 3 g cefazolin for 24 hours. Patients were kept in the hospital for 48 hours with further management being dictated by the clinical outcome. After delivery, chorionicity, preoperative diagnosis, and cord coagulation were confirmed by necropsy, pathologic examination, or both. Details of neonatal and pediatric outcome of the babies were obtained by our own pediatric unit (Figure 1a, 1b; Figure 2a, 2b).

Results

We performed bipolar cord coagulation in nine cases (Table 1) of monochorionic diamniotic twin (or triplet) pregnancies discordant for major fetal anomalies including anencephaly

Table 1. Clinical details of the cases

Case	Indication	Gestational age at procedure (week)	Complications	Gestational age at delivery (weeks)	Outcome
1	TRAP	18	-	38	A/W
2	TTTS	22	-	37	A/W
3	Hidrops fetalis	20	-	37	A/W
4	Triamniotic triplet discordant for anencephaly	18	-	34	A/W
5	Inencephaly	19	PROM	20	TOP
6	Anencephaly	22	-	36	A/W
7	Anencephaly	21	Preterm delivery	28+4	PND
8	TRAP	22	-	37	A/W
9	Lumbar meningomyelocele	20	-	38	A/W

TRAP: twin reversed arterial perfusion; A/W: alive and well; PROM: premature rupture of membranes; TOP: termination of pregnancy; PND: postnatal death.



Figure 1. Ultrasonographic image of the acardiac fetus at 18 weeks of gestation (a) and postnatal photograph (b).

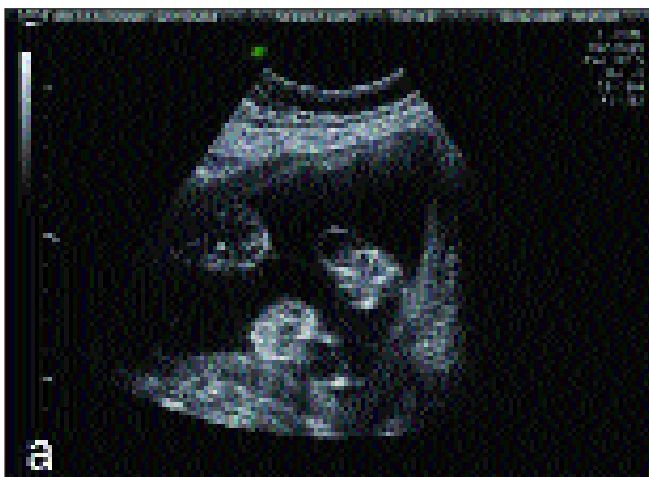


Figure 2. Monochorionic triamniotic triplet with co-triplet anencephalic fetus ultrasonographic image (a) and postnatal photography (b). Bipolar coagulation site of the umbilical cord (arrow).

(n=2), inencephaly (n=1), lumbar meningomyelocele (n=1), hydrops fetalis (n=1), acardiac twin (n=2) (Figure 1), twin-to-twin transfusion syndrome (stage III, n=1) and monochorionic triamniotic triplet discordant for anencephaly (n=1) (Figure 2). The procedure of bipolar coagulation was carried out in mean 20 min (R=15-40 min) without maternal complication. There were no failures of cord occlusion. The mean gestational age of the cases at intervention and at delivery were 20 weeks (R=16-24) and 34 weeks (R=20-38), respectively. One case was aborted at 20 weeks of gestation because of preterm premature rupture of membrane after the procedure. Another case was delivered before 32 weeks, at 28+4 week of gestation, and died six month postnatally for reasons unrelated to intervention. The other seven cases were delivered after 32 gestational weeks. In eight cases, the procedure were performed without fetal complication. Perinatal outcome of live birth was 88.8% (n=8/9). Postnatal ages of surviving seven babies were between 8-40 months, and they have been growing up as healthy.

Discussion

Selective termination of one fetus is a treatment option in well-selected cases of complicated monochorionic diamniotic twin pregnancies. Case selection, appropriate timing of the procedure, and choice of the optimal technique to be used remain uncertain. When the umbilical cord or major fetal vessels are subjected to obliteration, this should be effective and permanent to arrest fetofetal transfusion and to preclude postmortem interfetal hemorrhage. Early severe twin-to-twin transfusion syndrome, twin reversed arterial perfusion sequence, monochorionic diamniotic twin discordant for early severe intrauterine growth retardation, monochorionic diamniotic twin discordant for major anomalies and chromosomal anomalies are the indications (1,2).

Several techniques were reported for occluding the umbilical cord flow, such as Nd:YAG laser fetoscopic ligation, monopolar thermocoagulation, bipolar cord ligation and alcohol ablation (5-9). Laser and bipolar cord coagulation are the favored techniques in clinical practice.

Nd:YAG laser coagulation of the umbilical cord is attractive because it is a single-port procedure, and is fairly quick and easy. It has been done as early as 16 weeks but it has been shown to have a high failure rate after 20-22 weeks because of the larger size of the hydropic cord (1,2). The procedure may also be impaired when amniotic fluid is heavily stained, leading to poor transmission of light. This may be overcome by the use of amniocentesis (10). Along with preterm labor and rupture of the membranes, the risks of this procedure theoretically include vessel perforation (11).

Bipolar cord coagulation is a simpler and effective technique for cord ligation. The procedure can be carried out under ultrasound guidance using 2-3 mm bipolar forceps, which is inserted through an adapted cannula

[trochar]. It takes less time, can be carried out through a single port, and can also be performed successfully late in pregnancy up to 24-26 weeks. Coagulation is done at power settings of 25-50 W applied for 10 to 30s, and is repeated until the color Doppler confirms the absence of flow (12). Our experience comprises a series of nine consecutive cases treated by this technique (8 twin pregnancies and 1 sets of triplets). In one case the procedure was carried out for twin-to-twin transfusion syndrome, in two cases for twin reversed arterial perfusion sequence, and in six cases for structural anomaly (one of which was a triplet pregnancy).

In our initial series of nine cases, the survival rate was 88.8%. Lewi et al. reported a survival rate of 83% in a series of 80 consecutive cord occlusions performed by bipolar cord coagulation for various indications (13). Taylor and associates reported 40% of patients delivering before 32 weeks, the median interval from procedure to delivery being 10 weeks (range 0-20 weeks) and the median gestation at delivery being 33 weeks (range 24-38 weeks) (12). However, Lewi and colleagues, reported 21% of pregnancy delivering before 32 weeks and the median gestation at delivery as 35.4 weeks (range 24-40.4 weeks) (13). In our series, two patients (22%) delivered before 32 weeks; the median interval from procedure to delivery was 14 weeks (range 16-20 weeks), and the median gestation at delivery was 34 weeks (range 22-38 weeks).

All selective termination procedures risk incidental demise of the co-twin both at the time of the procedure and in the weeks following its completion. Procedure-related preterm labor, preterm delivery, preterm premature rupture of membranes, and placental or myometrial bleeding have all been described following selective fetal reduction (14). Iatrogenic preterm premature rupture of membranes, a term coined by Deprest and coworkers, remains the most common complication, occurring at rates 10-30% (10). Less frequent complications include placental abruption, chorioamnionitis, and amniotic band syndrome. Vasospasm of fetal vessels may lead to cessation of Doppler flow mimicking complete occlusion during the procedure, and subsequent devastating interfetal hemorrhage after completion of the procedure. Another potential risk is a large perforation of the separating membrane of a monochorionic diamniotic gestation, leading to both the live and reduced twin being in the same amniotic sac. This complication, like selective reduction in monochorionic monoamniotic pregnancy, can result in subsequent cord entanglement and death of the remaining healthy twin (13,15). In our cases, one patient had preterm premature rupture of membranes and underwent a termination. In the other eight cases, the procedure were performed without fetal complication.

In conclusion, ultrasonography-guided bipolar cord coagulation as a selective fetocide can be successfully

performed to increase the survival rate or prevent morbidity of healthy co-twin in the cases of monochorionic diamniotic twins (or triplets) discordant for major fetal anomalies candidate for selective termination, and should be offered as a choice of prenatal therapy after the parents were informed of the details of the intervention and the follow up.

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