

Acardiac twin pregnancy: successful intrauterine ablative treatment with alcohol at 14 weeks of gestation

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Summary

Twin reversed arterial perfusion (TRAP) sequence is a serious condition of monochorionic twin pregnancy, occurring in approximately one in 35,000 cases. First trimester treatment of TRAP sequence is controversial with higher incidence of procedure related complications. Present case demonstrates a TRAP sequence that was managed by intrauterine treatment with one-ml 100% pure alcohol injection into the abdominal part of the umbilical artery and obliteration of the acardiac twin at 14 weeks of gestation. Antenatal follow-up was uneventful and elective cesarean section was performed at 39 weeks' gestation. Postnatal outcome of the pump twin was excellent at 30 months after birth. Early second trimester elective ablation by alcohol injection can be an inexpensive, alternative, and reasonable minimal invasive treatment option to prevent fetal loss of pump twin before mid and late second trimester in perinatology centers where intrafetal cord occlusive methods are not available.

Key words: Acardiac twin; Twin reversed arterial perfusion sequence; Monochorionic pregnancy; Intrafetal therapy; Elective reduction; Alcohol.

Introduction

Twin reversed arterial perfusion (TRAP) sequence is a rare complication of monochorionic pregnancies that is characterized by the presence of a TRAP or acardiac twin perfused by an apparently normal, pump twin [1]. It is presented by lack of heart development associated with a spectrum of malformations and reduction anomalies in one of the twins, which is perfused in a paradoxical retrograde fashion by a structurally normal 'pump' twin through a single artery-to-artery anastomosis [2].

It is an extremely rare anomaly, occurring with a reported incidence of one in 35,000 deliveries, one in 100 monozygotic twins, and one in 30 monozygotic triplets [3, 4]. This condition is associated with high risk of perinatal death of the pump twin due to cardiac insufficiency and polyhydramnios induced preterm birth [5, 6]. The diagnosis of TRAP can be usually made by detection of a grossly abnormal co-twin within a monochorionic pair and showing the absence of head and cardiac pulsation in the acardiac twin. Paradoxical blood flow may be visualized by colour Doppler ultrasound to confirm the diagnosis. Once diagnosed, the primary aim of management is to improve survival chances for the structurally normal pump twin [5-8].

Several methods including cord occlusion techniques, or an intrafetal approach to ablate the vasculature in the acardiac twin have been proposed for intrauterine treatment

of acardiac twin pregnancy [9, 10]. Elective reduction of the acardiac twin at 12-14 weeks was reported as a minimally invasive and effective method to improve survival in a recent case study and meta-analysis [11]. In light of published literature, the authors aimed to discuss and present this case of acardiac twin in which obliteration of the cord using one-ml 100% pure alcohol injection into the abdominal part of the umbilical artery was performed and a good postnatal outcome for the pump twin was achieved.

Case Report

A 27-year-old, G1 P0 women was referred at 14 weeks' gestation with a suspicion of acardiac twin pregnancy. A monochorionic, diamniotic twin pregnancy was diagnosed; one twin demonstrated normal anatomic development with normal nuchal translucency measurement whereas the other one appeared amorphous without upper trunk and cardiac activity. Color Doppler ultrasound demonstrated reverse blood flow to the acardiac, acephalic fetus.

The patient was counseled about the risks of the pregnancy and possible complications of the treatment modalities. Selective termination of acardiac fetus was decided. Twenty-one gauge needle was oriented to intra-abdominal segment of umbilical artery of the acardiac fetus under continuous ultrasound guidance and one-ml 100% pure alcohol was applied from the catheter at 14 weeks' gestation. The procedure was terminated when cessation of blood flow was observed on color Doppler ultrasonography. Second trimester sonography and fetal echocardiography revealed normal anatomic surveillance for the pump twin.



Figure 1. — Photograph of post-cesarean specimen of the placenta at term with acardiac twin in a case of monochorionic, diamniotic TRAP sequence. Black arrow indicates the acardiac twin and ablated vessel.



Figure 2. — Isolated postmortem view of the elected reduced acardiac twin.

Antenatal follow-up was uneventful. Elective cesarean section was performed at 39 weeks of gestation by referring obstetrician. Photograph of post-cesarean specimen of the placenta at term with acardiac twin in present case of monoamniotic TRAP sequence and isolated postmortem view of the acardiac twin is demonstrated in Figure 1 and 2, respectively. The newborn physical examination findings were normal and the child had normal neurologic development at 30 months after birth.

Discussion

Acardiac twin is a rare congenital anomaly characterized by formation of a malformed fetus with an absent or non-functional rudimentary heart. The etiopathogenesis of this anomaly is abnormal single arterio-arterial placental vascular communication between the twins, leading to imbalance of interfetal circulation. Reversed blood flow in the umbilical artery of the acardiac twin causes atrophy of the heart and other organs [5, 6]. Perinatal mortality for the pump twin has been estimated to be up to 55%, while it is usually fatal for the acardiac twin [1].

Treatment by occlusion of this vascular connection has been proposed since the early 1980s [7]. As less invasive techniques improved, invasive surgical methods has been abandoned. Percutaneous ultrasound guided circulation occlusive injections have been attempted. Pure alcohol has been used as a vascular sclerosant agent [9-11]. Umbilical cord ligation, cord coagulation by laser, endoscopic intrafetal laser coagulation, and radiofrequency ablation are the treatment modalities used in the second trimester with high survival rates [9-12]. Even the median time of diagnosis of this abnormality are earlier as a consequence of first trimester nuchal translucency screening programs; intrauterine interventions have been performed after 16

weeks of gestation. The TRAP sequence can be treated by several approaches. Patients may be managed expectantly or prophylactic intervention in the first trimester or at 16-18 weeks may be scheduled [5]. Abdominal circumference ratio or twins weight ratios more than 50% and 70%, respectively, have been proposed as invasive intervention indications [1]. Evidence of congestive heart failure in the donor or hydrops of pump twin are poor prognostic factors for twin survival [1, 5, 9, 10] and these conditions should be considered as invasive intervention indications. Conservative management of TRAP sequence in which the acardiac fetus was equal or smaller than 50% of the weight of the pump has been reviewed as a safe option versus invasive treatment alternatives [13].

In present case, alcohol injection was used due to technical accessibility. Thrombosis of the umbilical vessel and transient bradycardia and fetal loss of the pump twin have been reported as a result of multicenter experience after the alcohol ablation [14]. Even the present authors' early second-trimester intervention resulted with a good perinatal outcome; intravascular transfers of the ablative chemical to the pump fetus may provoke these procedure-related complications. On the other hand, 60% of TRAP cases diagnosed at 11-14 weeks reached spontaneous arrest of the abnormal reverse blood flow and 61.1% of their pump twin died or suffered brain damage [11]. Chaveeva *et al.* concluded survival rate might be improved by elective intervention at 12-14 weeks as the authors performed in the present case. Tan *et al.* [9] identified 32 reports published until 2002 including 74 cases of TRAP sequences treated by minimally invasive technique, like cord occlusion and intrafetal ablation. The median gestational age at treatment was earlier, the median gestational age at delivery was later,

and the median treatment to delivery interval was longer in patients treated with intrafetal ablation. Intrafetal ablation was found to be associated with a lower rate of premature delivery or rupture of membranes before 32 weeks. They concluded that intrafetal ablation techniques are simpler, safer, and more effective when compared with cord occlusive methods [9]. Cohort study results for intrafetal laser treatment in TRAP sequence reported 82% pump twin survival rate [10]. Subgroup analysis demonstrated that adverse pregnancy outcome after laser treatment; intrauterine fetal demise and preterm delivery before 32 weeks of gestation was significantly lower when the procedure was performed before 16 weeks of gestation [10]. Recently successful result of high intensive ultrasound treatment for TRAP sequence has been reported as a non-invasive therapy [15].

In conclusion, as performed in the present case, early second-trimester elective ablation by alcohol injection can be an inexpensive alternative and reasonable minimal invasive treatment option for TRAP sequence in perinatology centers where intrafetal cord occlusive methods are not available.

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