

Case Report

INTRAFETAL LASER COAGULATION FOR TRAP SEQUENCE IN A COMPLICATED DICHORIONIC TRIAMNIOTIC TRIPLET PREGNANCY CASE REPORT AND REVIEW OF THE LITERATURE

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Summary

A rare case is reported of twin-reversed arterial perfusion (TRAP) sequence in a triplet pregnancy, fetal intervention in the first trimester and pregnancy outcome. We report a case of TRAP sequence complication in dichorionic triamniotic triplet pregnancy, with a normally developing fetus and an acardiac fetus connected via arterio-arterial anastomoses in a monochorionic diamniotic twin pair and a separate fetus. TRAP sequence was diagnosed at 13 weeks in triplet pregnancy after in vitro fertilization (IVF) and embryo transfer of two blastocysts. Color Doppler assessment showed persistent arterial flow in the acardiac twin. Intrafetal laser coagulation was carried out at the time of the diagnosis, and the pregnancy outcome was two survivals at 36.4 weeks of gestation.

Key words: twin reversed arterial perfusion (TRAP), fetal intervention, triplet pregnancy, intrafetal laser coagulation

Introduction

Twin-reversed arterial perfusion (TRAP) sequence is a rare complication of monochorionic twinning, affecting 1 in 35 000 pregnancies, and 1 in 30 monochorionic triplets [1]. The monozygotic twinning is a condition that now is becoming increasingly more common, and is seen mostly in vitro fertilization (IVF) pregnancies. The incidence of monozygotic twinning in natural conception is 0.4%, whereas in assisted conceptions it is five times higher [2, 3].

Monozygotic twin pregnancies have a different type of chorionicity, depending on the stage at which cleavage occurs to form the two embryos. If the cleavage occurs within three days after fertilization on the morula stage, the pregnancy will be monozygotic dichorionic. If the cleavage occurs after the 4th day but before the 8th day of fertilization, the pregnancy is defined as monochorionic diamniotic. When the blastocyst divides in the latest stage, the monozygotic pregnancy is monochorionic monoamniotic. Further splitting of the early embryo beyond the 13th day or after the formation of the embryonic disk is classified as a conjoined twin pregnancy, which is the fourth type of monozygotic pregnancy. The major impact on pregnancy outcome is associated with chorionicity

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rather than zygosity and the best time to determine the pregnancy as monochorionic or dichorionic is by a first trimester ultrasound examination [4]. The unique complications related to shared placentation are specific to monochorionic pregnancies, and the earliest type of abnormal vascular anastomoses between the fetuses presenting in the first trimester are found in twin reversed arterial perfusion sequence.

TRAP sequence is a formation of a structurally abnormal fetus with no heart activity connected via arterio-arterial anastomosis to its co-twin, acting as an “acardiac” twin and “pump” twin. The pathogenesis and the mechanism of blood perfusion of the acardiac malformation were first described by Van Allen et al. (1983) [5].

In all monochorionic pregnancies, there are shared vascular anastomoses found in the placenta. The imbalance of transfused blood among the fetuses can result in twin to twin transfusion syndrome (TTTS). The perinatal outcome in severe TTTS is related to a high percentage of perinatal deaths and neurological sequela before 28 weeks of gestation. Acardiac twin or twin reversed arterial perfusion sequence is the most extreme manifestation of TTTS and the specific vascular anastomoses linked to this condition are arterio-arterial, one vessel directly continuing into the other, allowing blood flow in both directions (bi-directional) with low resistance. Blood flows from an umbilical artery of the pump twin in a reversed direction into the umbilical artery of the acardiac twin via an arterio-arterial anastomosis, and then oxygen-depleted blood usually returns via a veno-venous anastomosis back to the pump twin, causing some degree of hypoxemia [5]. The reported outcome in pregnancies managed expectantly is a high rate of premature delivery due to polyhydramnios and death of the normal fetus caused by congestive cardiac failure [1].

Historically, the first attempts of intrauterine interventions were introduced with hysterectomy and removal of the acardiac fetus [6]. A recent meta-analysis has been carried out of the various techniques that have been used in the last 20 years for treatment of TRAP sequence to improve pregnancy outcome. These varied from the use of a coil insertion into the umbilical cord of the acardiac twin to umbilical cord ligation, laser coagulation of

placental anastomoses between the pump and acardiac twins, endoscopic laser coagulation and endoscopic or ultrasound-guided monopolar or bipolar coagulation of vessels within the cord of the acardiac twin. The interventions that were used recently were those of ultrasound-guided intrafetal coagulation performed by monopolar diathermy, laser or radiofrequency coagulation and alcohol injection. The most commonly used techniques were those of the cord bipolar coagulation, intrafetal laser coagulation and intrafetal radiofrequency and with each one of these techniques the survival rate of the pump twin was about 80%. Concerning intrafetal laser coagulation, the survival rate of the pump twin was 80%. The radiofrequency coagulation was performed at 20 weeks on the average, whereas intrafetal laser coagulation – at 16 weeks [7].

In the subsequent years, after a shift in ultrasound scanning to the first trimester with the introduction of a first trimester combined screening test for chromosomal abnormalities [8], there was an increasing number of cases with TRAP sequence diagnosed at 11-14 weeks. A new strategy of early intrauterine intervention was proposed, challenging the policy of treatment in such a condition in an attempt to avoid the hidden mortality of the pump twin between the 11-14 weeks and the scheduled procedure at 16-18 weeks [9].

Case Report

A 23-year-old woman was referred to our fetal medicine unit for diagnosis and treatment at 13 weeks of gestation. The pregnancy was achieved by in vitro fertilization with a transfer of two embryos at the blastocyst stage. The pregnancy was described as a triplet pregnancy with two fetuses with normal development for the gestational age and one fetus with no heart activity. At the time of the presentation, the medical history of the patient was unremarkable.

Ultrasound examination found dichorionic triamniotic triplet pregnancy complicated with TRAP sequence, a separate fetus with own placenta and amnion and monochorionic twins with one placenta and two amnions (Figure 1).

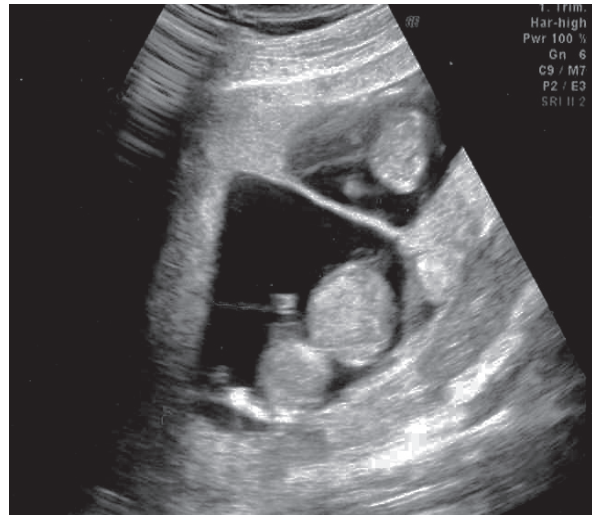
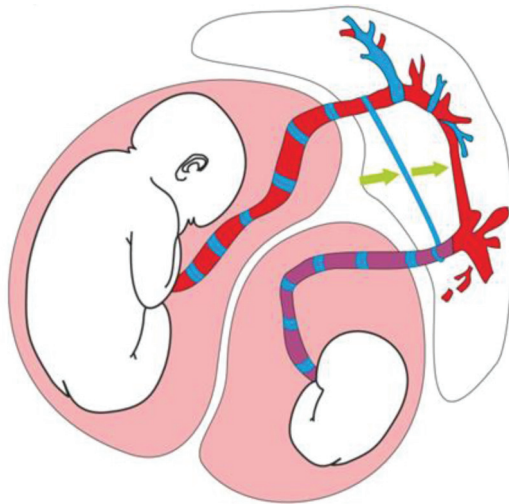


Figure 1. Twin reversed arterial perfusion sequence (TRAP)
Scheme of TRAP sequence with a normal fetus and an acardiac fetus connected via arterio-arterial anastomoses
TRAP sequence in dichorionic triamniotic triplet pregnancy

The measurements of the fetuses demonstrated two healthy fetuses with no visible fetal defects and signs of chromosomal abnormalities and an acardiac fetus with development of the lower

part of the body (acardius acephalus). Color Doppler demonstrated reversed arterial flow in the umbilical artery in the recipient acardiac twin (Figure 2).



Figure 2. Color Doppler in a TRAP sequence
Color Doppler, visualizing the iliac arteries of the acardiac twin.
Acardiac twin

As a routine practice in the first trimester, the performance of screening for chromosomal abnormalities included assessment of the blood flow in ductus venosus. There was a normal a-wave in the ductus venosus of the separate fetus and reversed flow in the ductus venosus of the donor twin, which may be interpreted as a marker of severe TTTS at this stage of the pregnancy, compatible with the findings of TRAP sequence [10].

After the diagnosis was made, extensive counseling with the parents, intrafetal laser coagulation of the feeding vessels of the acardiac twin was carried out. The patient was admitted for routine preoperative preparation, prophylactic antibiotics, and spasmolytic therapy. A written consent form was obtained, and the pregnancy details were included in a computerized hospital system.

Intrafetal laser coagulation

Intrafetal laser coagulation is a fetal intrauterine procedure performed by ultrasound guidance as described firstly by Nicolaidis et al. [7, 11, 12]. The technique we use was adopted from the Fetal Medicine Research Institute, King's College Hospital, London and was applied in the same manner in our department.

Intrafetal laser coagulation for treatment in TRAP sequence was carried out as previously described [7, 12, 13]. Ultrasound examination was performed to select the entry place of the maternal abdomen where the needle was inserted. Firstly, local anesthesia (10mL 1% lidocaine) was administered to the maternal skin, subcutaneous tissues and myometrium. Color Doppler was used to identify the vessels into the acardiac fetus. An 18 G needle (Cook Ireland Ltd., Limerick, Ireland) was introduced, and it was guided to the fetal pelvic vessels under continuous ultrasound visualization. A 400 µm laser fiber was pushed into the needle and advanced 1-2 mm beyond the tip of the needle to the selected point. Intrafetal laser coagulation was carried out with the use of NdYag-Laser (Dornier Med Tech, Wessling, Germany) with energy of 40 W. This resulted in cessation of blood flow within the acardiac twin and hyperechogenicity of tissues was obtained for 10 seconds. The duration of the procedure was approximately 10 min, and bedrest for the patient was recommended for at least one hour. An ultrasound examination on the same day confirmed that the intervention had gone uncomplicated. The heart activity in the pump twin was recorded for the medical documentation. The patient was discharged one day after the procedure and follow-up was undertaken at the local hospital. The patient reported that at 36.4 weeks of gestation there was a spontaneous rupture of membranes, and cesarean section was carried out on the same day with two survivals: a female and a male with birthweight 2350/2500 gr. There were no neonatal complications and the babies were discharged from the hospital in good health.

Discussion

Triplet pregnancies are at high risk of pregnancy

complications and have higher perinatal morbidity and mortality rates. In dichorionic triplet pregnancies managed expectantly, the incidence of miscarriage before 24 weeks of gestation is 9% and the rate of premature delivery after the 33rd gestation week is 39% [14-18].

The rarity of TRAP sequence complicating a triplet pregnancy raised the question of fetal interventions to improve pregnancy outcome. A review of 13 pregnancies complicated by an acardiac fetus managed expectantly showed poor pregnancy outcome with 10 cases miscarried or delivered before 32 weeks of gestation [19]. A meta-analysis reported a review of the literature with 28 cases of TRAP sequence in triplets with intrauterine interventions with survival of the pump twin in 78.6%, and an own series of expectantly managed pregnancies with survival of the normal fetus only in 42.9% [7].

The prognosis of the pump fetus and the separate fetus in a triplet pregnancy depend on the prematurity and, most specifically, on the condition of the pump twin that is likely to develop congestive heart failure. Early diagnosis and careful counseling are essential to provide the necessary management. The findings of a previous study have shown that this condition has exposed hidden mortality at between 11-13 and 16-18 gestation weeks, with a risk of death or brain damage in the pump twin in 60% of the cases [9]. The feasibility of fetal intervention in the first trimester for TRAP sequence in twins suggests that the management should be the same in multiple pregnancies. Early treatment by intrafetal laser has shown reduction of preterm labor and the earlier the procedure is performed, the higher the likelihood is of term delivery [7].

Conclusions

TRAP sequence in a triplet pregnancy is a rare condition. The prognosis and pregnancy outcome are closely related to early diagnosis in the first trimester and management with intrauterine intervention. The risk of hidden mortality of the pump twin between 12 and 16 weeks of gestation require timely therapy. Intrafetal laser coagulation is a feasible and effective method of treatment in the first trimester with good prognosis for the surviving fetuses.

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