

Spontaneous Cessation of Communicating Flow in a Twin Reversed-arterial Perfusion with Large Acardiac Twin

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We report a case of twin reversed-arterial perfusion diagnosed at the gestational age of 17 weeks. Though the acardiac/pump twin abdominal circumference ratio was 1.5, there was no critical Doppler finding of reverse A wave of ductus venosus, polyhydramnios or hydrops of the pump twin at the time of diagnosis, so the patient was regularly followed-up at clinic at 1-week intervals. Two weeks after the diagnosis, the patient requested surgical intervention because she was worried about the risk of heart failure in the pump twin. Fetoscopic guide laser coagulation of the communicating vessels was arranged. On the morning of the operation, cessation of the communicating flow was found, so the operation was cancelled. On the afternoon of the same day, amniocentesis was performed and the pump twin was found to have scalp edema. The patient was followed-up at clinic at 2-week intervals. A healthy female baby was born by vaginal delivery at the gestational age of 36 weeks.

KEY WORDS — twin reversed-arterial perfusion

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Introduction

Twin reversed-arterial perfusion (TRAP) sequence is a rare disorder that affects 1% of monozygotic twins [1]. Although the pump twin is normal structurally, expectant management is associated with a perinatal mortality rate of 35–55% [2,3]. The high mortality rate is largely a result of high-output heart failure or premature delivery caused by polyhydramnios and/or rapid growth of the acardiac twin. Risk factors that are associated with an increased likelihood of pregnancy loss include polyhydramnios, large acardiac twin (abdominal circumference [AC]

ratio of acardiac/pump twin size ratio > 50%), and preterm labor [3].

We report a case of TRAP with large acardiac twin, where the AC (acardiac/pump) ratio was 1.5 at 17 weeks of gestation. Fetoscopic surgery to coagulate the communicating vessels was arranged 2 weeks after diagnosis at the mother's request. On the morning of the scheduled surgery, however, the communicating flow was found to have ceased spontaneously. On the afternoon of the same day, the pump twin was found to have scalp edema. The pregnancy ended with vaginal delivery at 36 weeks of gestation. This case highlights the

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possibility of spontaneous cessation of communicating flow in TRAP even with large acardiac twin; scalp edema in the pump twin after spontaneous cessation of communicating flow does not indicate poor prognosis.

Case Report

A 30-year-old, gravida 3, para 1, woman presented to our clinic at 17 weeks of gestation; she was transferred from a local hospital with the diagnosis of acardiac twin. The diagnosis of TRAP was confirmed at our hospital by detecting the retrograde perfusion flow in the acardiac twin's umbilical artery (Fig. 1). The acardiac/pump twin AC ratio was 1.5 at this examination, but there was no polyhydramnios or sign of heart failure in the pump twin (Fig. 2). Due to the high possibility of heart failure in the pump twin because of the high acardiac/pump AC ratio, the patient was followed-up at clinic at 1-week intervals.

Two weeks after the first visit, the mother requested surgical intervention as she was worried about the possibility of heart failure in the pump twin. After explaining the risks of fetoscopic surgery to her, fetoscopic coagulation of the communicating vessels was arranged. On the morning of the operation, sonographic examination was performed to reassess the condition of the pump twin. Cessation

of communicating flow was found. The operation was cancelled and amniocentesis for karyotyping was suggested. During amniocentesis on the afternoon of the same day, the pump twin was found to have slight scalp edema without obvious ascites. The pump twin's scalp edema subsided the next day and the patient was discharged. The karyotype of the fetus was a normal 46XX.

The mother was followed-up at 2-week intervals in our hospital with an uneventful course. She was scheduled for cesarean section due to previous cesarean section. However, at 36 weeks of gestation, the acardiac twin was delivered via vagina at home (Fig. 3). On arrival at our delivery room, pelvic examination revealed a dilated cervix of 6 cm.

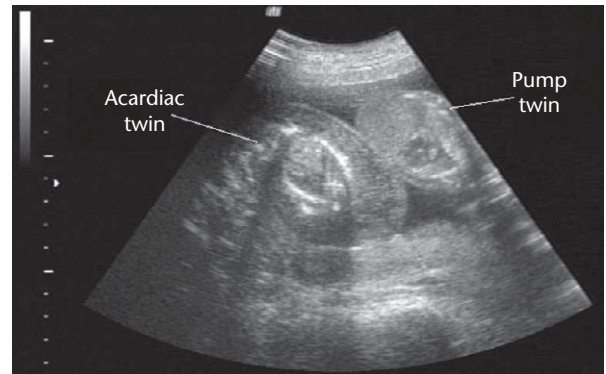


Fig. 2. The abdominal circumference of the acardiac twin is larger than that of the pump twin.

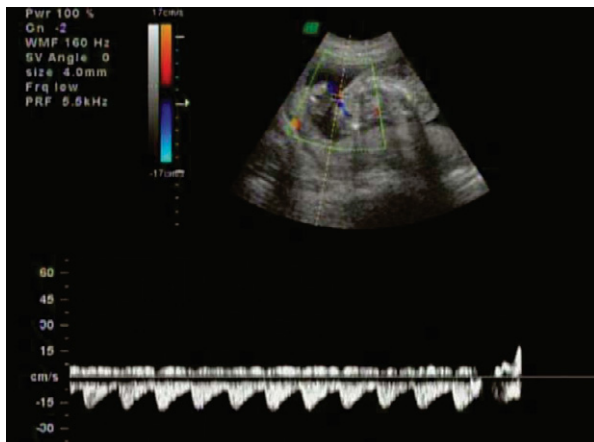


Fig. 1. Retrograde perfusion flow of the acardiac twin from the pump twin; the umbilical artery flow of the acardiac twin is retrogradely pumping into the acardiac twin.

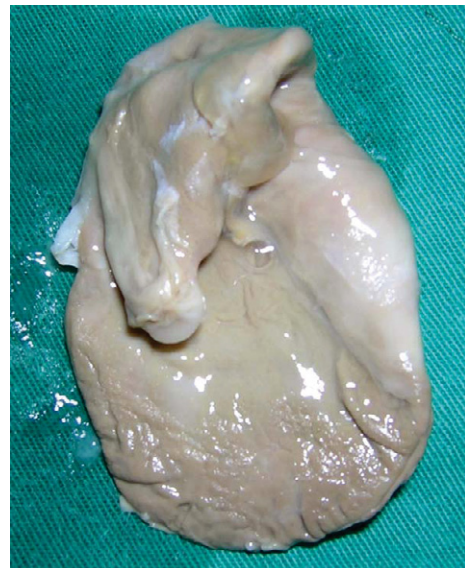


Fig. 3. The acardiac twin was delivered via vagina at home.

Vaginal delivery was thus suggested and a healthy girl weighing 2300 g was delivered smoothly. Post delivery, the brain and abdomen echo of the baby did not reveal any abnormal findings.

Discussion

Due to the rarity of TRAP, no standard management protocol is currently established [4]. Some favor a preventive procedure at an earlier gestational age before the occurrence of poor prognostic signs [5]. Some recognize that if poor prognostic signs are found, then invasive intervention is justified [4]. In a recent review paper, a classification system for the purpose of managing such cases was proposed [6]. According to this classification system [6] or to the intervention criteria of Quintero et al [4], our case should have undergone immediate intervention due to the high acardiac/pump AC ratio. But due to the invasiveness of fetal surgery, the fetal loss rate cannot be ignored. Preterm premature rupture of membranes before 34 weeks of gestation occurred in 18% of TRAP cases managed by fetoscopic surgery [5]. Thus, we did not suggest surgical intervention at the time of diagnosis.

It is difficult to perform a randomized trial on whether intervention for high-risk TRAP should be undertaken or not due to the low incidence of such cases. From our case, we learned that there is a possibility of spontaneous occlusion of communicating flow in TRAP even with high acardiac/pump AC ratio. Sullivan et al reported that four out of 10 cases of acardiac twins showed spontaneous cessation of flow to the acardiac twins, resulting in delivery of a healthy pump twin at term [7]. There was no Doppler index of the umbilical artery (S/D ratio) to predict the cessation of communicating flow [8], but torsion of the acardiac umbilical cord has been suspected to be the etiology of cessation of communicating flow [9]. Our case further demonstrates that the pump twin may suffer transient fluid overload manifesting as skin edema after the cessation of communicating flow. The mechanism

of transient pump twin edema after the cessation of communicating flow may be similar to the transient donor hydrops in twin-twin transfusion syndrome post fetoscopic surgery [10], and it is not a poor prognostic sign for the pump twin.

In conclusion, management options for TRAP should be individualized. Even with high acardiac/pump AC ratio, there is a possibility that the communicating flow will cease without intervention. After cessation of the communicating flow in TRAP, transient pump twin skin edema is not necessarily a poor prognostic sign.

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