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Case Report

Vaginal birth after caesarean section in a woman with twin reversed arterial perfusion sequence

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ABSTRACT

Twin reversed arterial perfusion (TRAP) sequence results from aberrant and asymmetrical twinning. It is a rare but serious complication of monozygotic twin pregnancies. It is characterized by a normally formed fetus that shows features of heart failure, and an acardiac twin. A 26-year-old pregnant woman, gravida 6, para 5, with mono-chorionic di-amniotic twin pregnancy at her 20th week of gestation, was diagnosed with TRAP sequence. Both the pump twin and the acardiac co-twin did not display any cardiac activity. Termination of pregnancy was accomplished by a successful vaginal birth after cesarean section (VBAC) following five prior cesarean sections. Proper and early diagnosis of TRAP sequence can aid in treatment selection and pump twin salvage. Induction of labor by both misoprostol and intracervical balloon catheter showed to be safe and effective in a woman with a uterine scar resulting from five prior cesarean sections.

Keywords: TRAP sequence, VBAC, Acardiac twin, Ultrasound

INTRODUCTION

Twin reversed arterial perfusion (TRAP) sequence is a unique and rare complication seen in monochorionic twin pregnancies.¹ It is recently increasing in incidence, reaching 2.6% of monozygotic twins, due to advancements in ultrasound detection and the widespread use of assisted reproductive technologies.² An aberrant placental vascular anastomosis during early stages of placentation elicits an unbalanced blood flow between the monochorionic twins, resulting in a normally developed fetus (also called the pump twin) shunting reversed deoxygenated blood into its co-twin. The latter becomes morphologically compromised and eventually acardiac and non-viable.³ The perinatal mortality of the pump twin reaches 55%, and it is mainly caused by congestive heart failure, polyhydramnios, preterm labor and delivery, and intrauterine growth restriction.^{4,5} In this article, we report

a case of successful vaginal birth after cesarean in a multiparous woman with a monochorionic twin pregnancy complicated by TRAP sequence, after documenting an absent fetal heart rate in the pump twin.

CASE REPORT

A 26-year-old pregnant woman, gravida 6, para 5, with a monochorionic twin gestation at her 20th week, was referred to our outpatient clinics for advanced ultrasonography examination. Earlier, she received a diagnosis of absent cardiac activity in one of her twins. Her medical and family histories were insignificant. She only reported a fourth degree consanguinity with her husband.

On ultrasonography, she was found to have an intrauterine monochorionic diamniotic twin pregnancy. Twin A appeared morphologically normal and its morphometric

measurements were commensurate with 18 weeks and 5 days of gestation, but no cardiac activity was detected (Figure 1). Twin B, however, was morphologically abnormal. Only a fetal head was apparent, and thus fetal measurements could not be taken. Similarly, it didn't show any cardiac activity (Figure 1). Under the light of these findings, TRAP sequence was considered in the radiologic differential diagnosis of this pregnancy.



Figure 1: Ultrasonography of the monochorionic diamniotic twins showing both the pump twin (right) and its acardiac co-twin (left).

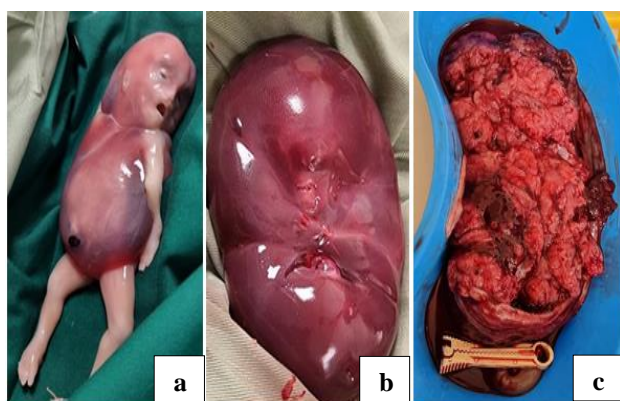


Figure 2: Images of the twins and the placenta (a) the morphologically normal pump twin, (b) the acardiac acormus twin, and (c) placenta.

The woman was then admitted to our labor and delivery (L&D) unit for a trial of labor. She received, according to our department protocol, half the misoprostol dosage suggested for fetal death by FIGO (2017) due to her previous cesarean scar.⁶ She received 100 mcg of misoprostol vaginally every 6 hours. Cervical ripening was enhanced by the insertion of an intracervical balloon catheter. She had an uncomplicated delivery of her twins (Figure 2a and b). The first twin was morphologically normal and developed. The second twin showed only a fetal head without trunk or extremities. Placental separation and delivery occurred within 10 min after

delivery of the second twin. Gross inspection showed a single placenta without obvious vascular anastomosis (Figure 2c). A bedside ultrasound was then performed and it excluded any uterine scan rupture or retained products of conception. She was discharged home one day postpartum in stable conditions.

DISCUSSION

TRAP sequence should be considered when an ultrasound examination of a monochorionic twin pregnancy reveals morphological abnormalities in one of the fetuses.¹ Prenatal diagnosis can be made at the end of the first trimester by detecting a rudimentary non-functional fetal heart (pseudocardiac), or absence of fetal heart (halocardiac), or by revealing placental vascular anastomosis using color Doppler which may show pathognomonic features of reverse flow on an artery-artery anastomosis.⁷ Early diagnosis allows determining the possible therapeutic interventions, as well as timely termination of pregnancies that have a high mortality rate in the pump fetus.⁸

Halocardia is morphologically divided into four subgroups: acephalic (without cranial structures), anceps (with rudimentary cranial structures), acormus (has cranial structures but lacking body or extremities) and amorphous (non-recognizable).⁹ Acardia acormus, as in our case, is encountered in only 10% of acardia cases which emphasizes the rarity of our case findings.

Intrauterine fetal demise of a morphologically abnormal twin resembles an acardiac fetus, and hence ultrasonography plays an important role in differentiating TRAP sequence from intrauterine fetal demise.¹⁰ The ultrasonographic findings of continued fetal growth and persistent intra-fetal blood flow raise the suspicion of TRAP sequence over fetal demise.¹

The optimal management of an acardiac twin pregnancy is still debated as it is rarely encountered.¹¹ The goal of therapy is the salvage of the pump twin, although it has a high perinatal mortality rate reaching 55%.^{4,11} Some authors suggest invasive methods that aim to discontinue blood flow to the acardiac twin. These methods include endoscopic umbilical cord ligation, thermocoagulation of the umbilical cord and aorta under ultrasonographic guidance, sclerosis of the umbilical cord with alcohol, and coagulation of the umbilical cord.¹²⁻¹⁵ However, Suvillain et al suggested that conservative follow-up had a lower mortality rate compared to invasive methods.¹⁶ In their series, a live birth of the pump twin occurred in 9 out of the 10 women, and 4 of the total 10 cases resolved spontaneously with the cessation of the blood flow to the acardiac twin after 19 weeks of gestation.

In our case, the pump twin was lost at the first ultrasound and thus, no therapeutic intervention could salvage it. The only possible management was to terminate the pregnancy. In a study by Sansregret et al, they concluded that a trial of

labor in women with twin pregnancies and a prior cesarean section had a shorter hospital stay and that there were no differences in maternal morbidity between births by trials of labor and by elective cesarean section.¹⁷ Induction of labor was then performed according to the FIGO recommendations for misoprostol administration in case of fetal death. FIGO recommends abiding to the department protocol in case of prior cesarean scar, so we administered half the recommended dosage.⁶ An intracervical balloon catheter was inserted to fasten the induction process as current guidelines recommend using it to promote cervical ripening in women with prior cesarean who require induction of labor.¹⁸

To date, the use of vaginal misoprostol for intrauterine fetal demise in the context of a prior uterine scar remains debated. A case report published in 2021 by Beshar et al suggested that misoprostol can be a reasonable option in case of fetal demise in a woman with a uterine scar.¹⁹ Yet, induction of labor in such women carries a 1-5% risk of uterine rupture.²⁰ Fortunately, our patient had a successful VBAC without any evidence of uterine rupture.

CONCLUSION

TRAP sequence is a rare complication of monochorionic twin pregnancies. Proper and early diagnosis by ultrasound, with the help of Doppler, can aid in treatment selection and pump twin salvage. In case of fetal demise, induction of labor by both misoprostol and intracervical balloon catheter showed to be safe and effective in a woman with a uterine scar resulting from five prior cesarean sections.

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