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Case Report

Unveiling a hidden trap: a case of unexpected acardiac twin diagnosed at cesarean section

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ABSTRACT

Twin reversed arterial perfusion (TRAP) sequence is a rare complication of monochorionic twin pregnancies, occurring in 1 in 35,000 births. It is characterized by retrograde arterial perfusion of a malformed acardiac twin by the structurally normal pump twin through placental vascular anastomoses. A 29-year-old primigravida with a monochorionic diamniotic twin pregnancy was diagnosed with a viable Twin A and intrauterine foetal demise of Twin B on early ultrasonography. Serial scans showed progressive anomalies in Twin B, including a large anechoic cranial cyst, overlapping skull bones and diffuse subcutaneous edema. At 28 weeks of gestation, the patient presented with preterm premature rupture of membranes. An emergency caesarean section was performed for cord presentation of Twin A. A 770-gram male twin were delivered and required intensive neonatal support but died on day 2 of life due to multiorgan failure, birth asphyxia and very low birth weight. The acardiac twin weighed 2390 grams with well-formed lower limbs but severely malformed upper body structures. TRAP sequence results from deep placental arterial-to-arterial anastomoses causing reversed blood flow and variable maldevelopment of the acardiac twin. Pump twins, though anatomically normal, are at significant risk of cardiac overload and poor perinatal outcome, especially when the acardiac twin exhibits disproportionately large mass. Minimally invasive foetal therapies such as radiofrequency ablation and high-intensity focused ultrasound have shown improved survival in selected pregnancies. TRAP sequence remains a rare but serious complication with high morbidity and mortality. Early detection, multidisciplinary counselling and consideration of foetal therapy are crucial for optimizing pump twin outcomes.

Keywords: TRAP sequence, Acardiac twin, Pump twin, Monochorionic twins, Fetal therapy

INTRODUCTION

Twin reversed arterial perfusion (TRAP) sequence is a rare condition that occurs exclusively in monochorionic monozygotic twin pregnancies and carries a poor prognosis. It has an incidence of approximately 1 in 35,000 births and occurs in 1 out of every 100 monozygotic twin pregnancies. In TRAP sequence, one fetus is nonviable with multiple anomalies and an underdeveloped or absent heart, while the other—known as the pump twin—supplies blood to the acardiac twin through abnormal placental vascular connections.[1]

In the acardiac twin, although body and limb movements may be present, the absence of a detectable heart on ultrasonography (USG) and the identification of vascular anastomoses in the placenta via Doppler USG help confirm the diagnosis prenatally. The mortality rate for the acardiac twin is 100%, while the pump twin faces a mortality rate of approximately 50%, primarily due to

heart failure and, in some cases associated with polyhydramnios.

CASE REPORT

An informed written consent of the patient and her relative for publication and use of their case details was obtained on 15/04/2025. A 29-year-old primigravida, registered and with regular antenatal visits, diagnosed as a monochorionic diamniotic (MCDA) twin gestation with foetal demise of Twin B, presented to the labour room with complaints of leaking per vaginum at 28 weeks of gestation.

Ultrasonography at 14 weeks of gestation revealed a viable intrauterine Twin A in breech presentation with cardiac pulsations (147 bpm), weighing 152 grams with parameters of 16 weeks of gestation. Twin B was deformed with oedema and a completely anechoic area within the calvarium replacing the brain tissue.

An obstetric growth scan at 18 weeks showed a single live intrauterine Twin A in variable presentation with cardiac pulsations (147 bpm), weighing 239.81 grams. Twin B demonstrated deformation with diffuse subcutaneous oedema suggestive of the "space suit sign." The patient was advised a detailed anomaly scan for Twin B at a tertiary healthcare centre.

A second-trimester anomaly scan performed at the tertiary centre suggested a monochorionic diamniotic pregnancy with no congenital anomaly in Twin A (weighing 408 grams with parameters of 20 weeks of gestation). Twin B had a large anechoic cyst within the calvarium, overlapping skull bones with Spalding sign, mild kyphosis of the foetal spine, and dense echoes with cystic areas in the amniotic sac.

The patient was subsequently registered at the tertiary centre at 18 weeks after referral due to absent cardiac activity in Twin B. On admission, her vitals were stable. The uterus was over-distended, foetal heart sounds were not localized, and there were no uterine contractions. Per vaginal examination revealed an uneffaced cervix with os admitting the tip of a finger and frank leaking.

An ultrasound was repeated as foetal heart sounds were not localized with a handheld Doppler. The scan showed Twin A, closer to the OS, in breech presentation with foetal heart rate of 150 bpm, weighing 875 grams with parameters of 25+6 weeks, and oligohydramnios with AFI <5. Doppler study showed raised PI in the umbilical artery (1.9). PI in the MCA could not be evaluated. PI in the left uterine artery was 0.8 and in the right was 1.2. Twin B, located away from the OS, had a large anechoic cyst within the calvarium, overlapping skull bones, and dense echoes with cystic spaces within the amniotic fluid.

The patient and relatives were counselled regarding the poor prognosis of Twin A and the high risks associated with the pregnancy. Two doses of injection betamethasone were administered 24 hours apart as steroid cover, and injectable antibiotics were started.

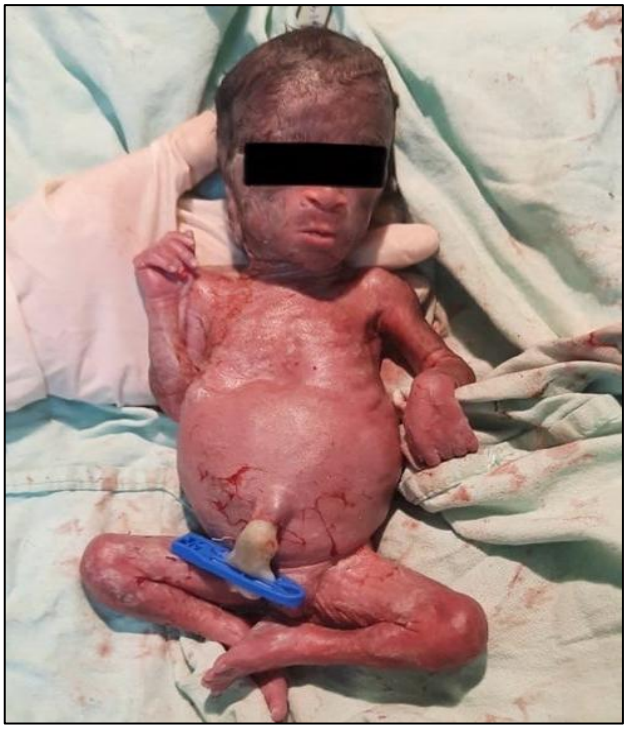

Figure 1: Pump twin (twin A) delivered at 28 weeks of gestation, weighing 770 grams.

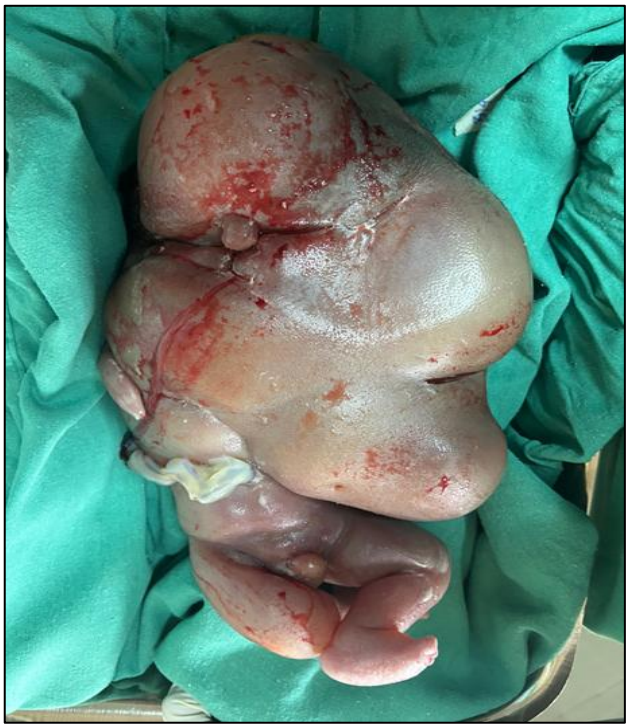

Figure 2: Acardiac twin (twin B) weighing 2390 grams, showing well-formed lower limbs and an amorphous cephalic mass.

After 24 hours, per vaginal examination showed a 30% effaced cervix, one-finger loose dilation, absent membranes, and face presentation of Twin A. Fetal heart rate was monitored hourly. After 48 hours, cervical os was 3 cm dilated, 50% effaced, and cord pulsations were felt. An emergency lower segment caesarean section was performed for cord presentation. Twin A, a male baby weighing 770 grams, in transverse lie, was delivered by breech extraction and cried after stimulation (Figure 1). Twin B (acardiac twin) was delivered two minutes later, weighing 2390 grams with well-formed lower limbs and genitalia, poorly formed upper limbs, and an amorphous mass with cephalic elements (Figure 2). The placenta was monochorionic monoamniotic (Figure 3).

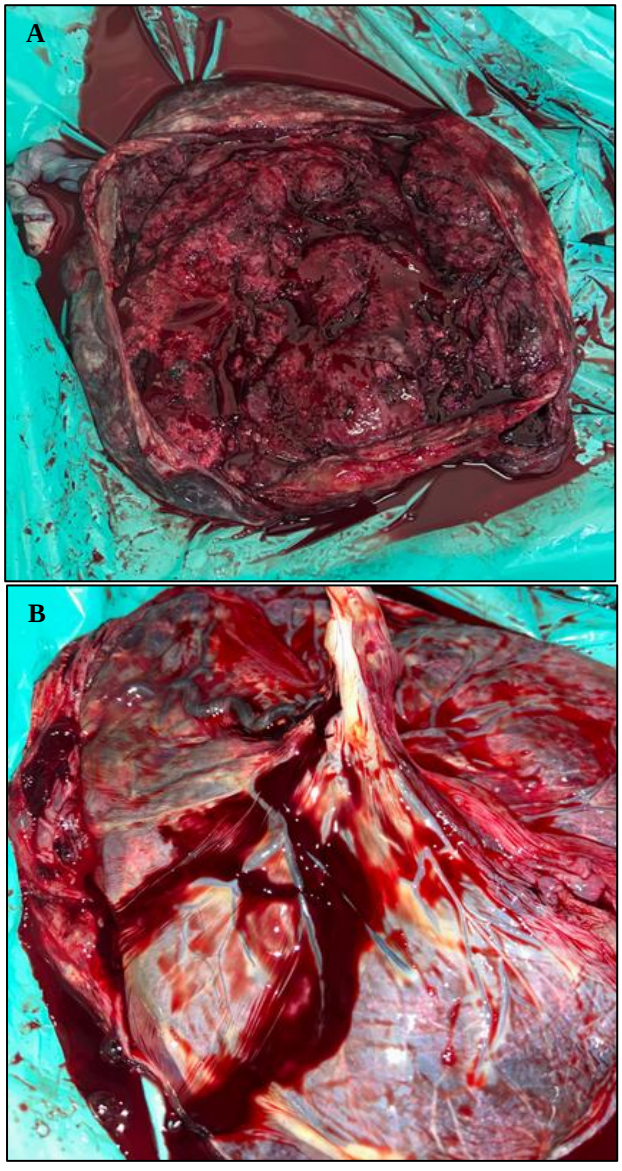

Figure 3 (A and B): Monochorionic monoamniotic placenta with short umbilical cord.

The pump twin had no visible malformations. No obvious placental anomaly was noted. The umbilical cord of the acardiac twin was short. There were no intraoperative complications, and the postnatal period was uneventful for the mother. The pump twin was shifted to the neonatal ICU, intubated, and transfused with fresh frozen plasma and blood. The baby succumbed on the second day of life due to multiorgan failure syndrome, birth asphyxia, respiratory distress syndrome, and very low birth weight.

DISCUSSION

Acardiac twinning, historically known as chorioangiopagus parasiticus, represents the most extreme manifestation of TRAP sequence.[2] TRAP is classified according to the degree of cephalic and truncal maldevelopment. The pathogenesis involves malformation in the acardiac twin due to deep placental anastomoses in early embryogenesis. TRAP sequence was defined by Van Allen et al. in 1983.[3]

These vascular anastomoses may be vein-to-vein, artery-to-artery, or between the two umbilical cords through the chorionic plate. Deoxygenated blood flows from an umbilical artery of the pump twin to that of the acardiac twin in a reversed direction, as the perfusion pressure of the pump twin overwhelms that of the recipient. This deoxygenated blood preferentially reaches the iliac vessels, perfusing only the lower body and disrupting normal development of upper body structures.

The acardiac twin is classified into four types based on morphological anomalies[4]:

Acardius acephalus

Most common; head and upper extremities unformed.

Acardius anceps

Cranial structures and neural tissue partially developed; trunk and upper extremities present.

Acardius acormus

Cephalic structure present but no trunk; umbilical cord inserted into the head; rarest type.

Acardius amorphous

Most severe malformation; no distinguishable cephalic or truncal structures.

Pump twins usually have a normal karyotype, whereas recipient twins may have chromosomal anomalies.[5] Pump twins are at risk for congestive heart failure and hydrops due to the burden of perfusing the parasitic twin.

Although pump twins are typically anatomically normal, their survival rate is approximately 50% due to cardiac complications or prematurity.[3] In monochorionic twins with one twin IUFD on first-trimester ultrasound, TRAP sequence should be considered in the differential

diagnosis.[6] Ultrasound surveillance for foetal weight ratio, cardiac failure, and polyhydramnios is crucial. Estimated foetal weight can be calculated by comparing abdominal circumference ratios or using the prolate ellipsoid formula (length × abdominal diameter ÷ 2).[7]

Conservative management has an excellent prognosis when the estimated fetal weight of the acardiac twin is less than one-fourth of the pump twin. The risk increases to 100% when the acardiac twin weighs more than 70% of the pump twin.[8] Radiofrequency ablation (RFA) is the preferred treatment, with improved outcomes. The North American Fetal Therapy Network reviewed 98 cases between 1998 and 2008 in which RFA was performed; median gestational age at delivery was 37 weeks and 80% of donor neonates survived.[8]

High-intensity focused ultrasound (HIFU) is a non-invasive therapeutic tool that causes biological reactions at the target site. Non-thermal cavitational effects are defined as the effects of the physical forces of ultrasound waves on micro-gas bubbles within the tissue. The characteristic compression and rarefaction by cavitation cause micro-gas bubbles in the tissue to contract and expand. These rapid changes may cause damage to and around the cell. In a study of six cases conducted by Seo et al, the total survival rate of pump fetuses 2 years after HIFU was 67% and the efficiency rate (the proportion of cases with occlusion or reduced blood flow on ultrasound after HIFU) was 83%.[10] After more than 2 years of follow-up, the surviving infants had no severe clinical complications and no postnatal developmental problems. HIFU can reduce the cardiac load of the pump fetus and, as it does not require uterine puncture for fetal therapy, there were no fatal complications, such as bleeding, rupture of membranes or infection. Thus, HIFU therapy may represent a less-invasive treatment for TRAP sequence in early pregnancy.[10]

CONCLUSION

TRAP sequence is a rare yet severe complication of monochorionic twin pregnancies with high perinatal morbidity and mortality. Early diagnosis, timely referral to a tertiary centre, and close surveillance of the pump twin are crucial. Advancements such as radiofrequency ablation and high-intensity focused ultrasound offer promising outcomes.

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