

**Case Report**

# **Twin Reversed Arterial Perfusion Sequence (TRAPS): A Case Report**

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**ABSTRACT**

**Introduction and importance:** *Acardiac twins are a complication of monozygotic twin pregnancies. Twin Reversed Arterial Perfusion Sequence (TRAPS) is reported to occur in approximately 1% of monochorionic twin pregnancies and 1 in 35,000 pregnancies. The objective is to provide a brief description of acardiac twins.*

**Presentation of case:** *A 25-year-old multigravida, 35/36 weeks of twin pregnancy. On ultrasound examination of acardiac twins, no heart structure, head bones, and sternum were found, and the shape of the fetus was unclear. Vaginal delivery was carried out; the first live female baby was born, followed by the acardiac baby. The acardiac baby is seen to have imperfect development of the head and upper extremities, Spina bifida, deformity of the lower extremities, Phocomelia of the lower limb, and Split foot. After 9 days, the baby's condition improved, and she was allowed to go home.*

**Discussion:** *TRAPS's etiology and pathophysiological mechanisms are still poorly understood. Ultrasound findings show an absent or non-functioning heart in one twin, malformation with cystic hygroma, and generalized edema. The first baby (donor twin) was born prematurely, weighing 2200 grams, and there were no signs of heart failure, while the second baby (acardiac) was born weighing 1050 grams. Current therapy for acardiac twins aims to prevent congestive*

*heart failure in the donor twin and stop blood flow to the acardiac twin. **Conclusions:** Early diagnosis of abnormalities in monochorionic pregnancy using ultrasound in the first trimester plays a role in assessing the prognosis for the viability of the pregnancy.*

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## 1. INTRODUCTION

The number of twin pregnancies continues to increase to 33.7 per 1000 births in 2013.<sup>1</sup> Perinatal mortality in twin pregnancies is five times higher than in singleton pregnancies.<sup>2</sup> Twin Reversed Arterial Perfusion Sequence (TRAPS) or acardiac twins are reported to occur in approximately 1% of monochorionic twin pregnancies and 1 in 35,000 pregnancies.<sup>2</sup> The use of first-trimester ultrasound (USG) examinations and assisted reproductive technology play a role in increasing the incidence of twin pregnancies, including monochorionic twins.<sup>3</sup> A 2015 study estimated the incidence of acardiac twins to be 2.6% of monochorionic twin pregnancies and 1 in 9,500 to 11,000 pregnancies.<sup>3</sup> TRAPS is caused by abnormal placental vascular anastomoses that cause increased arterial pressure in 1 twin and backflow to the other twin. As a result, the donor fetus (pump twin) pumps low-oxygen blood to the recipient fetus (acardiac fetus).<sup>1</sup> Furthermore, the recipient fetus experiences severe hypoxia, and its limbs are not fully formed. The acardiac fetus only has the lower body or only tissue mass. Therefore, the mortality of the acardiac fetus is 100%. The donor fetus may experience congestive heart failure and hydropic changes due to the continued pumping of blood to the acardiac fetus.<sup>2</sup> Mortality for the donor fetus is 50-75%. Until now, there has been controversy regarding the management of TRAPS.<sup>3</sup> This case report aims to provide a brief description of acardiac twins and how early diagnosis using ultrasound in the first trimester plays a role in assessing the prognosis for the viability of the pregnancy.

## 2. CASE PRESENTATION

We report a case, a 25-year-old multigravida, 35/36 weeks of twin pregnancy, who was admitted to the Department of Emergency Obstetrics and Gynecology with signs of labor. The patient underwent antenatal care two times at the primary health care center, two times at the hospital, and once at the fetal subspecialist. She denied any family history of congenital malformations. She had no history of radiation exposure or adverse medication use during early pregnancy.

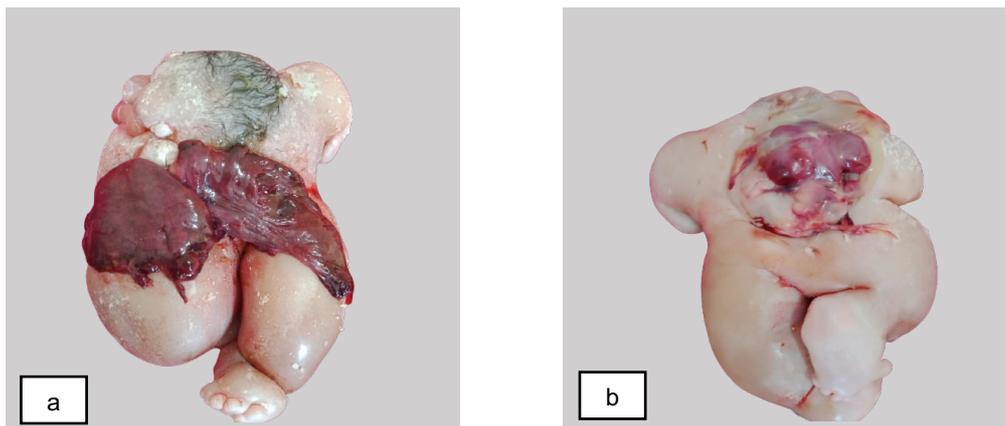
On physical examination, the vital signs were typical: the fundal height was 39 cm, the abdominal circumference was 87 cm, the estimated external fetal weight was 3393 grams, and the fetal heart rate was 142 bpm. The results of the vaginal examination showed a cervical opening of 4 cm with protruding amniotic fluid. No abnormality was detected in laboratory examinations. Vaginal delivery was performed; the first baby girl was born alive, birth weight of 2200 grams, a birth length of 46 cm, and an Apgar score of 7/9, followed 30 minutes later by the second baby (acardiac) with a birth weight of 1050 grams. The first twin looked normal without external abnormalities and was admitted to the Neonatal Intensive Care Unit (NICU) (Fig 1). The acardiac baby had imperfect development of the head and upper limbs, spina bifida, deformity of the lower limbs, Phocomelia of lower limbs, and split foot (Fig 2a and 2b).

There were two amnions and one placenta with two umbilical cords. The results of the ultrasound examination of the acardiac twin fetus did not find the structure of the heart, skull, or sternum, and the shape of the fetus was unclear, like a mass (Fig 3). Umbilical artery Doppler studies showed similar indices for both fetuses, and there was no reversed flow. A plan to manage her expectantly was made by two weekly ultrasound scans and Doppler velocimetry. The patient was counseled on the risks of preterm birth, cardiac failure, and higher perinatal mortality associated with the condition. However, the patient did not come for the next appointment and lost to follow-up. She was then presented with signs of labor at 35/36 gestational weeks.

The patient was transferred to the ward in good condition and discharged from the hospital on the second day postpartum. After 9 days of being treated in the NICU, the baby's condition improved, and she was allowed to go home with a weight of 2000 grams. Follow-up of the baby until the age of 1 year showed no complications. Weight, growth, and development are appropriate for her age.



**Fig. 1.** The Donor twin was born with no external abnormalities, a birth weight of 2200 grams, a birth length of 46 cm, and an Apgar score of 7/9.



**Fig. 2.** The Acardiac twin was born with a birth weight of 1050 grams. a) The front view showed imperfect development of the head and upper limbs and Phocomelia of lower limbs (b) The Back view showed spina bifida and split foot.



**Fig. 3.** Ultrasound of acardiac twin with no structure of the heart, skull, or sternum, and the shape of the fetus was unclear, like a mass.

### 3. DISCUSSION

TRAPS is a rare occurrence in monozygotic twins. Therefore, it should be suspected in monochorionic pregnancies. The etiology and pathophysiological mechanisms of TRAPS still need to be better understood. Two hypotheses have been put forward; the first hypothesis states an arterial-artery and venous-venous anastomosis in the monochorionic placenta. The pressure flow in the placental artery of one twin exceeds the other and causes reverse flow in the donor twin. As a result, the donor fetus (pump twin) pumps low-oxygen blood to the recipient fetus (acardiac fetus), and the caudal segment perfusion is relatively more significant than the cranial segment. This condition can cause secondary disorders in organogenesis due to lack of oxygen. The second hypothesis states that the formation of an abnormal heart early during embryogenesis initiates the development of this anomaly.<sup>4</sup>

Ultrasonography (USG) is a safe and non-invasive procedure for detecting malformations. USG can be used to predict fetal prognosis and assist in providing counseling to the mother. Currently, the diagnosis of acardiac twins is based solely on USG. USG findings show an absent or non-functioning heart in one twin, malformation with cystic hygroma, and generalized edema.<sup>5</sup> In this case, no heartbeat can be found on the USG examination, and the heart structure cannot be identified.

In this case report, the acardiac baby was found to have imperfect head and upper limb growth, spina bifida abnormality, which occurred due to the failure of the neural tube to close entirely in the first trimester, there Phocomelia of lower limb abnormality, namely there was no partial or complete thigh and lower leg. However, there was a foot and split foot due to the absence of partial or complete middle toe and metatarsal (figures 2 and 3). There were two amnions and one placenta with two umbilical cords. The donor twin was treated in the NICU, and after 9 days, the baby's condition improved, and she was allowed to go home weighing 2000 grams.

The perinatal mortality ratio in donor twins reaches 50-75%, which is caused by complications such as congestive heart failure, polyhydramnios, prematurity, and even intrauterine fetal death.<sup>5</sup> Studies showed that prognosis depends on the ratio of donor and acardiac fetal weight; if the estimated weight of the acardiac twin is less than a quarter of the donor twin, the prognosis is excellent without further therapy. In this

patient, the weight of the acardiac twin cannot be calculated using standard formulas based on ultrasound biometry (such as Hadlock's) because its anatomical structure is incomplete, so it has been proposed to use the following formula:  $(1.2 \times \text{longest length}^2) - (1.7 \times \text{most extended length})$ .<sup>6</sup>

The risk of congestive heart failure increases in the third trimester because the acardiac twin becomes more prominent in size, and the burden on the donor twin's heart becomes heavier.<sup>5</sup> Another thing that can increase the risk of congestive heart failure to 94% in the donor twin is if the acardiac twin's weight reaches more than half the size of the donor twin.<sup>6</sup> In this case, the first baby (donor twin) was born prematurely, weighing 2200 grams, and there were no signs of heart failure, while the second baby (acardiac) was born weighing 1050 grams.

Current therapy for acardiac twins aims to prevent congestive heart failure in the donor twin and stop blood flow to the acardiac twin. Treatment options include observation with close supervision (conservative therapy) and surgical intervention. Invasive intervention methods are through umbilical cord occlusion techniques or intrafetal ablation, which can be performed above 16 weeks of gestation.<sup>5</sup> Occlusion techniques have been tried with embolization, ligation, laser coagulation, bipolar diathermy, and monopolar diathermy, while intrafetal ablation is performed with alcohol, monopolar diathermy, laser, and radiofrequency.<sup>7</sup> Research shows intrafetal ablation is more straightforward, safer, and effective than umbilical cord occlusion techniques.<sup>7</sup> Nevertheless, these procedures for in-utero therapy are available at a small number of institutions in developing countries such as Indonesia.

#### **4. CONCLUSION**

TRAPS is a rare complication in monochorionic pregnancy. In this case, one twin has no heart, while the other twin may have congestive heart failure and death. From this report, early diagnosis of abnormalities in monochorionic pregnancy using ultrasound in the first trimester is essential to see the prognosis of the viability of the pregnancy. Nevertheless, these procedures for in-utero therapy are available at a small number of institutions in developing countries such as Indonesia.

#### **ETHICAL APPROVAL**

Not Applicable.

#### **CONSENT FOR PUBLICATION**

All the authors have consented to the publication of identifiable details, including photographs, case history, and details within the text, which will be published in the above journal.

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### **Conflict of Interest Statement:**

The author declares that the case report was conducted without any commercial or financial relationships that could be construed as a potential conflict of interest.

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