Case Report

Monochorionic Triplet Pregnancy with an Acardiac Twin: A Poor Prognostic Case

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Abstract

Introduction: Twin Reversed Arterial Perfusion (TRAP) sequence, or acardiac twin, is a rare complication in monochorionic pregnancies due to abnormal placental anastomoses, leading to reversed perfusion and increased risk of heart failure, polyhydramnions, prematurity, and high mortality until 75% if untreated.

Case report: A 29-year-old multigravida with a monozygotic triamniotic triplet pregnancy at 20–21 weeks presented with vaginal bleeding. Ultrasound revealed two non-viable fetuses and one suspected intrauterine demise. Vaginal delivery produced two stillbirth females neonatus (295 g and 275 g) and one malformed acardiac fetus (160 g). The placenta was monochorionic with two umbilical cords. Final diagnoses: complete abortion, monozygotic triamniotic triplet pregnancy, and acardiac twin (TRAP).

Conclusion: TRAP in monochorionic triplet pregnancy carries poor prognosis. Early detection and referral to tertiary centers with fetal therapy are vital to improve outcomes.

Keywords: Acardius amorphous type; fetal demise; monochorionic triplet; TRAP sequence

Kehamilan Triplet Monokorionik dengan Janin Akardiakus: Kasus dengan Prognosis Buruk

Abstrak

Pendahuluan: *Twin Reversed Arterial Perfusion* (TRAP) *sequence* atau janin akardiakus merupakan komplikasi langka pada kehamilan monokorionik yang terjadi akibat adanya anastomosis plasenta abnormal. Kondisi ini menyebabkan aliran perfusi terbalik sehingga meningkatkan risiko gagal jantung pada janin donor (*pump twin*), polihidramnion, prematuritas, serta mortalitas tinggi hingga mencapai 75% bila tidak ditatalaksana.

Laporan Kasus: Seorang wanita berusia 29 tahun, multigravida, dengan kehamilan triplet monozigot triamniotik pada usia kehamilan 20 – 21 minggu datang dengan keluhan perdarahan pervaginam. Pemeriksaan ultrasonografi menunjukkan dua janin tidak hidup dan satu janin diduga mengalami kematian intrauterin. Persalinan pervaginam menghasilkan dua neonatus perempuan lahir mati (berat 295 g dan 275 g) serta satu janin akardiakus dengan malformasi (berat 160 g). Plasenta monokorionik dengan dua tali pusat. Diagnosis akhir: abortus komplit, kehamilan triplet monozigot triamniotik, dan janin akardiakus (TRAP *sequence*).

Kesimpulan: TRAP pada kehamilan triplet monokorionik memiliki prognosis yang buruk. Deteksi dini dan rujukan ke pusat pelayanan tersier dengan fasilitas terapi fetal sangat diperlukan untuk memperbaiki luaran kehamilan.

Kata kunci: Janin akardiakus tipe amorfus; kematian janin; TRAP *sequence*; triplet monokorionik

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Introduction

Twin Reversed Arterial Perfusion (TRAP) sequence is a rare complication unique to monochorionic multiple gestations, with an estimated incidence of 1% of monochorionic twins or 1:35,000 births.[1] It arises from the presence of large arterio-arterial and veno-venous anastomoses in a monochorionic placenta. In this pathophysiology, a developmentally normal "pump" twin perfuses the parasitic acardiac twin in a reversed direction. Deoxygenated blood is shunted from the pump twin to the acardiac twin via an artery-to-artery anastomosis, leading to the recipient twin's severe malformations, including acardia, acephalus, and limb deficiencies.[2] The pump twin is jeopardized by the high-output cardiac state required to support the parasitic circulation, leading to a mortality risk of 50 – 75% due to cardiac failure, hydrops, and polyhydramnios-induced preterm birth.[3]

Risk factors for the TRAP sequence are primarily related to monochorionic placentation itself. Advanced maternal age, assisted reproductive technologies (which increase the rate of multiple gestations), and a history of monozygotic twinning may indirectly increase the risk. The key prognostic factor is the acardiac twin's weight relative to the pump twin's estimated weight; a ratio exceeding 50% is associated with a significantly higher risk of pump twin compromise.[4,5]

Prenatal diagnosis is achieved through ultrasonography, demonstrating a nonviable fetus with absent cardiac activity and disordered morphology. Doppler studies confirming reversed flow in the umbilical artery of the acardiac twin are pathognomonic.[6] Management strategies range from expectant monitoring to invasive fetal therapies. Fetoscopic laser coagulation of the acardiac twin's umbilical cord has emerged as the gold standard treatment in viable pregnancies, effectively interrupting the parasitic circulation and significantly improving pump twin survival rates.[6] Research conducted by Anca FA *et al.* found that fetoscopy to block the umbilical cord in acardiac twins resulted in normal fetal development.[7] This case report discusses a monochorionic triplet pregnancy complicated by acardiac twins, resulting in mid-trimester delivery, and reviews the poor prognosis if not managed early.

Case Report

A 29-year-old multigravida was referred to our tertiary care hospital at an estimated gestational age of 20 – 21 weeks with a diagnosis of triplet pregnancy, threatened preterm labor, and one suspected intrauterine fetal demise (IUFD). The patient's chief complaint was vaginal spotting and mucus discharge for five days before admission. She denied any other associated symptoms, such as fever, abdominal pain, or rupture of membranes. There was no known family history of congenital malformations or multiple gestations.

Vital signs on admission were as follows: Blood Pressure 100/70 mmHg, Heart Rate 115 bpm, Respiratory Rate 20 breaths/min, SpO_2 99% on room air, and Temperature 36.5°C. Obstetric examination revealed: Leopold I: soft uterus, fundal height 22 cm; Leopold II: fetal back on the left; Fetal Heart Rate: 158-164 bpm; a hard, round mass was palpated on the right lower quadrant; Leopold III: Ballottement was positive; Leopold IV: Convergent. Laboratory investigations revealed a hemoglobin level of 10.8 g/dL and a mild leukocytosis of 10,300/µL.

The patient subsequently underwent a spontaneous vaginal delivery. The first baby was a stillborn female with a birth weight of 295 grams. The second baby was a stillborn female weighing 275 grams. The third delivered product was a poorly formed, non-

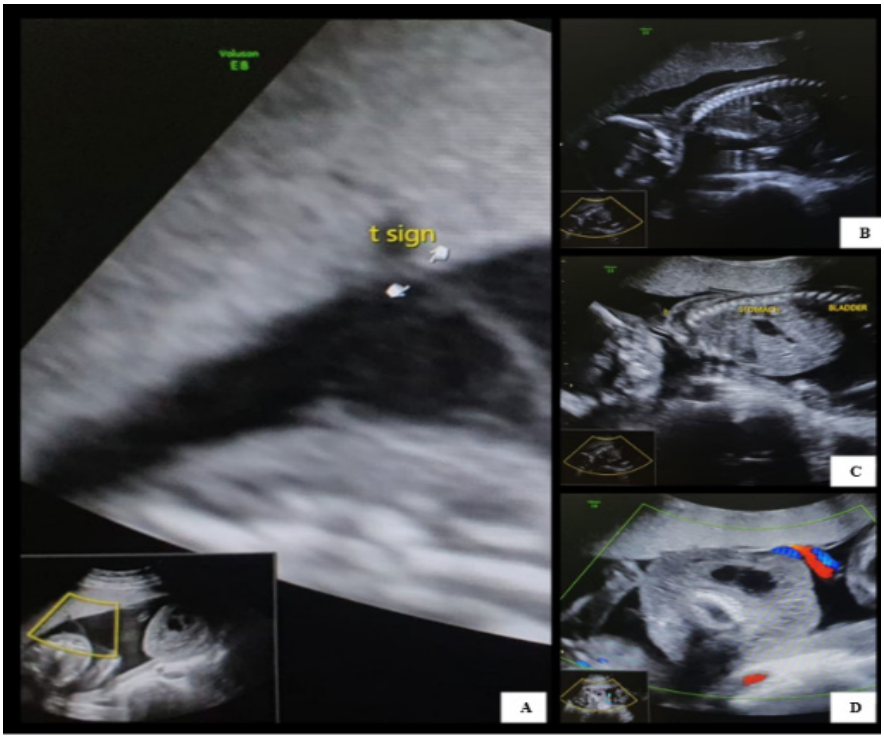

Figure 1 (A) Showing T-Sign, (B) First Fetus with Left Hydronephrosis, (C) Second Fetus with Intraventricular Septal Defect, and (D) Third Fetus

viable fetus weighing 160 grams, consistent with an acardiac twin. Examination of the delivered placenta revealed a single monochorionic disk with two umbilical cords, one supplying the two presumably monozygotic live-born twins and the other attached to the acardiac mass.

The final diagnoses for this case were: Complete Abortion + G2P1A0 at 20 - 21 weeks of gestation, triplet pregnancy, monochorionic triamniotic, acardiac twin (suspected TRAP sequence), suspected VSD in twin II, and left hydronephrosis in twin I. This condition can be differentiated from fetus papyraceus and severe fetal anomalies associated with cardiac arrest.

The patient was managed with inpatient care to allow close maternal and fetal monitoring, including regular assessment of vital signs, uterine contractions, fetal heart rate, vaginal examination findings, and signs of labor progression. Non-pharmacological management consisted of expectant management with a plan for vaginal delivery while maintaining strict observation. Pharmacological therapy included oral micronized progesterone (Microgest 200 mg twice daily) to support uterine quiescence and reduce the risk of preterm labor, nifedipine 10 mg three times daily as a tocolytic agent to suppress uterine contractions, folic acid supplementation (Folamil Genio once daily) to support maternal and fetal nutritional needs, and calcium supplementation (2 × 500 mg daily) to meet increased gestational requirements. The patient was also counseled to report worsening abdominal pain, with escalation to further obstetric intervention if contractions intensified, and a follow-up fetomaternal ultrasound was planned to reassess fetal status and guide ongoing management.

Discussion

This case exemplifies the poor prognosis

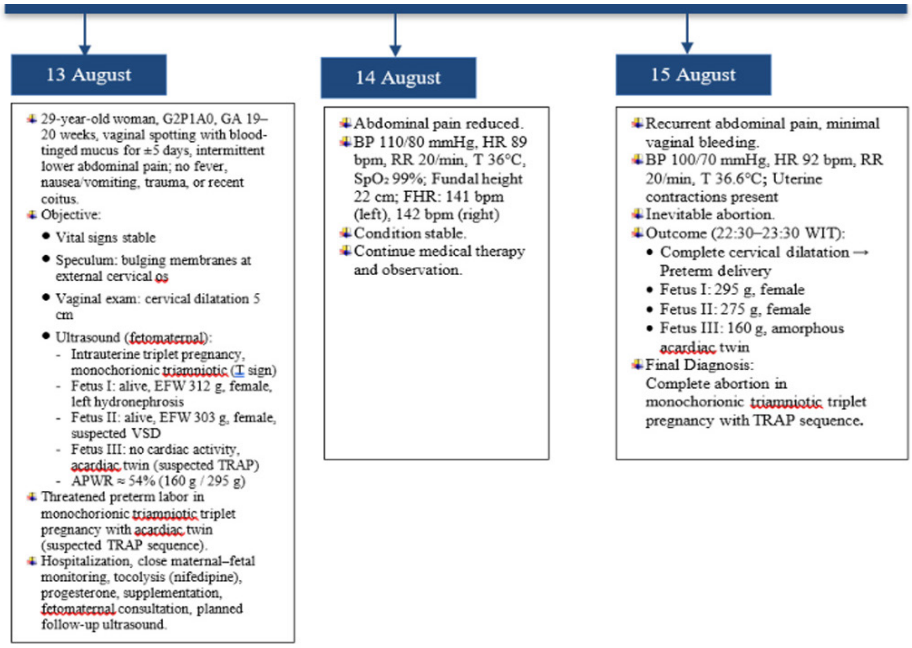

Figure 2 Timeline of Case

associated with TRAP sequence in a monochorionic triplet pregnancy, culminating in stillborn infant. The acardiac twin, perfused retrograde by its pump co-twin(s), exhibited classic severe malformations. The acardiac twin was perfused by the pump twin through arterio-arterial placental anastomoses, leading to significant cardiac strain on the pump twin. This increased hemodynamic load ultimately contributed to the development of polyhydramnios and the subsequent preterm labor and delivery at a non-viable gestational age. The acardiac twin exhibited severe maldevelopment, a hallmark of TRAP sequence, where reversed arterial flow from the pump twin supplies the acardiac mass with deoxygenated blood, typically resulting in absent upper body structures, acephalus, and acardia.[8,9]

The main risk factor for acardiac twinning or TRAP sequence is monochorionic placentation, which allows the formation of abnormal arterial anastomoses between twins. Pregnancies with monochorionic placentas often result from monozygotic twinning, which is known to be significantly increased in pregnancies resulting from assisted reproductive technology (ART). Therefore, ART is an indirect risk factor for TRAP, as it increases the likelihood of multiple and monozygotic pregnancies. In addition, in cases of monochorionic triplet pregnancies, the complexity of placental hemodynamics is greater, thus worsening the workload of the healthy fetal heart. Morphological factors such as the weight ratio of the acardiac twin to the pump twin are also important prognostic factors; the greater this ratio, especially if >50%, the higher the risk of heart failure and death of the healthy fetus. However, external factors such as maternal age, medical history, or lifestyle are not direct triggers of TRAP.[5,10]

The definitive diagnosis of TRAP is established by Doppler ultrasound, which characteristically shows reversed, retrograde flow in the umbilical artery of the acardiac twin.[11] In the present case, the initial suspicion was an intrauterine fetal demise (IUFD) of one triplet.

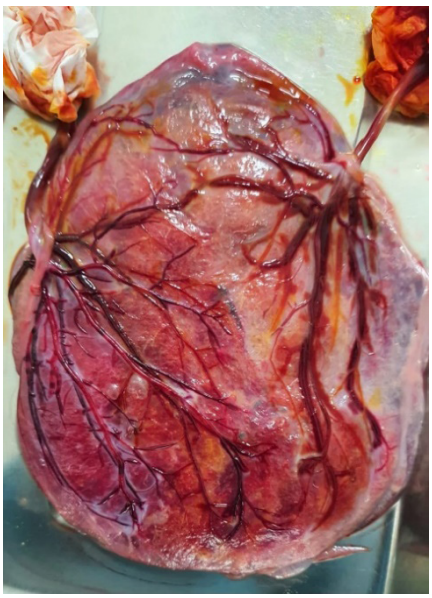

Figure 2 The Two Red Arrows Show The Umbilical Cords, The Yellow Line Dividing The Two Placentas Show The Equator Line, and The Red Circle Shows The Vascular Anastomosis

This is a common diagnostic pitfall, as the anomalous acardiac mass can be mistaken for a macerated fetus. A detailed Doppler study is, therefore, imperative in any monochorionic pregnancy with a demised or anomalously formed fetus to rule out TRAP sequence. Fetal demise in TRAP sequence represents a diagnostic and prognostic continuum rather than a single event. In early gestation, the acardiac twin may mimic intrauterine fetal demise (IUFD) due to the absence of cardiac activity, gross malformations, and reduced fetal movements. However, unlike true fetal demise, the acardiac mass may continue to enlarge due to persistent retrograde perfusion from the pump twin. This diagnostic pitfall has been well documented in recent literature, emphasizing that any suspected fetal demise in a monochorionic pregnancy must be evaluated with color and spectral Doppler to exclude ongoing vascular flow, which is pathognomonic for TRAP sequence. Failure to recognize this entity may result in delayed intervention and subsequent demise of the pump twin due to progressive high-output cardiac failure.[10,11]

Accurate determination of chorionicity is therefore critical in preventing misdiagnosis. One of the most reliable first-trimester sonographic markers of monochorionic placentation is the T sign, which represents the perpendicular insertion of the thin intertwin membrane directly into the placenta without intervening chorionic tissue. The T sign contrasts with the lambda (twin peak) sign seen in dichorionic pregnancies and reflects a shared placental circulation, predisposing to vascular ana

stomoses and complications such as TRAP sequence. Recent studies emphasize that early identification of the T sign allows timely surveillance, targeted Doppler assessment, and early referral to fetal therapy centers, thereby reducing the risk of pump twin demise that may otherwise be misinterpreted as spontaneous fetal loss.[12,13]

A key prognostic factor for pump twin survival is the acardiac-to-pump twin weight ratio (APWR). An APWR exceeding 50% is associated with a markedly higher risk of cardiac failure in the pump twin and poor obstetric outcomes.[14]

Another study conducted by Olutoye AS *et al.* found a ratio of acardiac-to-pump weight ratio (APWR) of 67% with poor prognosis in both fetuses.[15] In this case, the APWR was calculated to be approximately 54% (160 g/295 g), which indicated a severe prognosis and a high risk of pump twin decompensation, which manifested as preterm labor.

Abnormal fetal heart rate in TRAP sequence reflects progressive cardiovascular decompensation of the pump twin. Chronic low-resistance arterio-arterial placental shunting imposes a high-output circulatory state, initially resulting in compensatory tachycardia to maintain systemic perfusion.

As myocardial strain worsens, ventricular dysfunction develops, leading to reduced heart rate variability and, in advanced stages, bradycardia preceding fetal demise. Thus, abnormal fetal heart rate represents a late marker of pump twin cardiac failure and is associated with poor prognosis.[10,11]

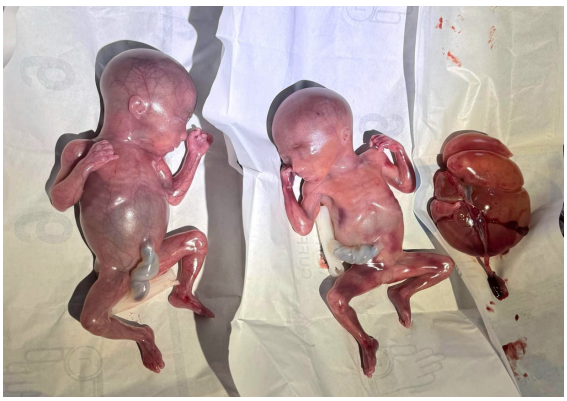

Figure 3 The Red Arrow Shows The First Female Infant (295 Grams), The Blue Arrow Shows The Second Female Infant (275 Grams), and The Yellow Arrow Shows The Third Infant, Not Yet Formed or an Acardiac Twin of The Amorphous Type (160 Grams)

TRAP sequence in acardiac twins is classified based on the degree of body development. Acardius acephalus is the most common form, characterized by the absence of a head but with partial development of the body and lower limbs. Acardius amorphous is the most severe form, appearing as an unshaped mass with minimal tissue and no clear body structures. Acardius acormus is defined by the presence of only a head without a body. Acardius myelacephalus is rare, with a formed head and partial spine but absent lower body. This classification is important as the degree of malformation correlates with the risk of complications for the pump twin.[16] In this case, the type of acardiac twin encountered was acardius amorphous.

The diagnosis of an acardiac twin consistent with a suspected twin reversed arterial perfusion (TRAP) sequence was established based on characteristic ultrasonographic findings. Differential diagnoses included fetus papyraceus and severe fetal anomalies with cardiac arrest. Fetus papyraceus results from early intrauterine fetal demise in multiple pregnancies and is characterized by absent vascular perfusion and fetal compression. In contrast, the affected twin in this case showed preserved somatic development despite absent cardiac activity, indicating ongoing perfusion.[17] Severe fetal anomalies with cardiac arrest may also present with absent cardiac activity; however, these conditions are not associated with reversed arterial perfusion. The presence of monochorionic placentation and reversed arterial flow on Doppler ultrasonography, a hallmark of TRAP sequence, supports the diagnosis and distinguishes it from other causes of fetal demise.[18,19]

Management options for TRAP sequence are contingent on gestational age, the APWR, and the clinical status of the pump twin. The Role of Fetoscopic Laser Ablation. Early prenatal diagnosis is the cornerstone of management. Upon confirmation of TRAP sequence, careful surveillance for signs of pump twin compromise is mandatory. In cases where the acardiac mass is large or signs of cardiac strain appear, fetal intervention is indicated to protect the pump twin. Fetoscopic laser coagulation of the acardiac twin's umbilical cord is the definitive treatment. This minimally invasive procedure aims to occlude the aberrant vascular communication, immediately relieving the hemodynamic burden on the pump twin.[15] After ablation, it's essential to conduct regular monitoring of both the pump twin and the acardiac twin. Ultrasound examination is performed to confirm cessation of blood flow to the

acardiac twin and to assess the well-being of the pump twin. The acardiac twin is left to regress, and if no complications arise, the pregnancy is continued until the appropriate time for delivery.[16]

Recent literature demonstrates high success rates with this approach. A 2022 systematic review by Buca *et al.*[20] reported survival rates exceeding 80-90% for pump twins following laser ablation therapy. The procedure is typically performed between 16-26 weeks of gestation and requires a highly skilled team at a tertiary care center. In this reported case, the late presentation with advanced cervical changes precluded any possibility of fetal intervention. The outcome underscores the natural history of the untreated TRAP sequence. The availability of fetoscopic laser ablation could have potentially altered the tragic outcome by allowing the pregnancy to progress to a more viable gestational age. This condition can be differentiated from fetus papyraceus and severe fetal anomalies associated with cardiac arrest based on ultrasonographic findings, including the presence of a monochorionic placentation, reversed arterial perfusion, and the absence of cardiac activity with preserved somatic development in the acardiac twin. This case highlights a rare presentation of TRAP sequence in a triplet pregnancy and underscores the diagnostic pitfall of misclassifying acardiac twin as IUFD. However, the delayed diagnosis and absence of fetal intervention limit the ability to assess the potential impact of timely treatment on perinatal outcomes.

Conclusion

Monochorionic triplet pregnancy complicated by an acardiac twin (TRAP sequence) carries a grave prognosis, often resulting in extreme prematurity and neonatal loss, as demonstrated in this case. Early prenatal diagnosis through detailed ultrasound and Doppler is paramount for timely intervention. Fetoscopic laser ablation of the acardiac twin's umbilical cord is a highly effective treatment that can significantly improve pump twin survival. The limited global availability of these advanced techniques necessitates improved early screening and referral systems to prevent the poor outcomes associated with this devastating condition.

Conflicts of Interest

The authors have no conflict of interest to declare.

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