

Twin Reversed Arterial Perfusion (TRAP): A Case Report

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Abstract

Background: Twin reversed arterial perfusion (TRAP) sequence is a rare and severe complication of monochorionic twin pregnancies. It represents the most extreme form of twin-to-twin transfusion syndrome and is characterized by retrograde arterial perfusion of an acardiac twin by a structurally normal “pump” twin. This abnormal vascular arrangement places the pump twin at significant risk of cardiac failure and perinatal mortality. We report a rare case of acardiac acephalic twin managed expectantly in a resource-limited setting.

Materials and Methods: This is a case report of a 28-year-old primigravida who presented at 25 weeks’ gestation with suspected fetal malformation. Clinical evaluation, laboratory investigations, and serial ultrasonography with amniotic fluid index (AFI) and estimated fetal weight (EFW) measurements were performed. A diagnosis of monochorionic twin gestation with one normal fetus and an acardiac acephalic twin was made. Due to unavailability of fetal interventional therapy, conservative management with close maternal and fetal monitoring was adopted.

Results: Serial ultrasonography showed stable growth of the pump twin with gradually declining AFI. The patient remained clinically stable until spontaneous rupture of membranes at 30 weeks + 3 days, necessitating emergency caesarean section. A live female neonate weighing 800 g was delivered with good Apgar scores, alongside an acardiac acephalic twin weighing 3.0 kg. The pump twin survived the immediate neonatal period.

Conclusion and Recommendations

Expectant management of TRAP sequence may result in favorable outcomes when the acardiac twin constitutes a relatively small proportion of the pump twin’s weight and there are no signs of cardiac failure. Early diagnosis, serial ultrasound surveillance, and timely delivery are essential, particularly in settings where fetal intervention is unavailable. This study aims to highlight the rare condition and how it presents on the ultrasound.

Keywords

Twin reversed arterial perfusion (TRAP); Acardiac twin; Monochorionic twins; Ultrasound

Introduction

Twin reversed arterial perfusion (TRAP) sequence, also known as *Chorioangiophagus parasiticus* represents a complex congenital anomaly in monozygotic multiple gestations and is the most extreme manifestation of the fetofetal transfusion syndrome.¹

The condition is extremely rare and account for 1:35,000 live births with an average risk of 1% among monozygotic twins.^{2,3}

Although the aetiology is unclear, it has been hypothesized that in a monochorionic twins gestation, the affected twin has no direct vascular connection to the placenta, but rather obtains all of its blood supply through an arterio-arterial anastomoses from the unaffected twins. Hence, blood perfused by haemodynamically advantaged twin (‘pump’ twin) to the other twin (recipient twin) by means of retrograde flow through its umbilical artery or arteries and returns through its umbilical vein.⁴ As a result of this anomalous connection, the recipient twin becomes acardiac and acephalic due to perfusion of deoxygenated blood by the donor twin who is also at

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risk of cardiac failure because of its expanded cardiac demand.^{5,6} The mortality rates without treatment could be as high as 50-70%.⁷ We hereby report this rare case of acardiac acephalic second twin resulting from TRAP.

Case Report

Mrs. F.M. is a 28-year-old primigravida whose last menstrual period was on 12th November, 2010. She presented in our facility at 25 weeks gestation with an ultrasound report done elsewhere; four weeks earlier, which indicated that she has a congenitally malformed foetus.

She has had no prior significant past medical or surgical history, except that she was just recently diagnosed as HIV positive prior to her presentation here. There was no positive history of twinning in her family and no history of drug use in the index pregnancy except the usual haematenics of folic acid and iron tablets. She had not formally registered for antenatal care and only presented for check up in the previous facility.

Mrs. F.M. was not ill looking on examination. Essential clinical findings were a weight of 84.5 kg and a normal blood pressure. Abdomen was grossly but uniformly enlarged (symphysio-fundal height of 48cm), with a firm and rather tensed feeling on palpation and not compatible with her estimated gestational age. No foetal heart sounds were heard on auscultation.

She was subsequently investigated. Complete blood count showed marginal anaemia with a 30% haematocrit and an elevated erythrocyte sedimentation rate (ESR) of 105mm/hour. Repeat HIV screen using Combi III was positive and CD 4 count was 321 cells/ml. Liver function test revealed a slightly elevated aspartate transaminase AST of 66 iu/L(0-49), electrolyte ,urea and creatinine were essentially normal.

The ultrasound done in this facility stated twin gestation, first twin normal active and with regular foetal cardiac pulsations (See Figure 1). Second twin is acardiac. The acardiac twin is seen as a stationary mass of placenta - like tissue. There is an increased liquor volume in the normal twin with an amniotic fluid index (AFI) of 16cm (See Figure 2) and female gender. The acardiac twin AFI is 4.7cm (See Figure 3 and 4). The conclusion was twin gestation, a normal at 25weeks+3d gestation and an acardiac acephalus twin.

The diagnosis of TRAP (Twin reverse arterial perfusion sequence) was made. The condition, its risks, management options including foetal image surgical intervention (FISI) and the prognosis of the condition including the outcome of the management for the surviving twin were extensively explained to the couple by the management team.

Management plan: Since FISI was not available in Nigeria in addition to the cost implications, the couple elected to have an expectant approach to the management. We decided to take up the challenge by monitoring all her laboratory parameters and checking the heart rate and rhythm and AFI weekly on outpatient basis and then twice weekly from 28 weeks on admission so as to identify when there is evidence of heart failure in the “pump” or surviving twin.

The plan also involved commencing antiretroviral drugs, HAARTS even with the challenge posed by the AST level while correcting the complete blood picture. The patient was expected to carry the pregnancy to about 32 weeks barring any complications (hospital admission was commenced at 28 weeks for closer monitoring) following which an elective caesarean delivery will then be effected since neonatal survival is assured. In the interim, parenteral dexamethasone therapy was commenced for 48 hours in weekly courses from the 28th week till delivery.

Serial measurements of AFI, estimated fetal weight (EFW) and ESR are as follows:

Parameters	26 weeks	27 weeks	28 weeks	29 weeks	30 weeks
Amniotic fluid index(cm)	14.0	12.5	11.6 /11.2	10.0 / 11.3	10.5 / 9.7
ESR(mm/hr)	95			132	105
EFW (g)	826	915	1056	1084	1089
Symphysiofundal height(cm)	48	46	44	44	43



Fig. 1: showing 3D sonogram image of the normal twin



Fig. 2: showing polyhydramnios associated with the pregnancy



Fig. 3: showing acardiac second twin with polyhydramnios

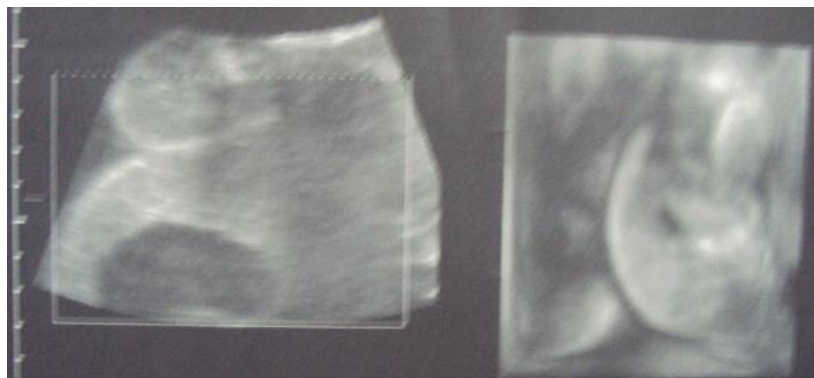


Fig. 4: showing ambiguous acardiac acephalic second twin



Fig. 5: Showing delivered ambiguous acardiac acephalic second twin.

Patient remained stable throughout admission for about 16 days until she spontaneously ruptured her membranes at 30 weeks+3days gestation necessitating an emergency classical caesarean section to be performed.

The findings were: first twin was live female 800g neonate with Apgar Scores of 6/1, 7/5 and 9/10. The acardiac twin was delivered by conversion of the

transverse lower uterine segment incision to a midline longitudinal incision (classical incision). A large solid and firm lifeless rounded mass with no head weighing 3.0 kg covered with skin and with upper limb appendages and twisted lower limbs with no identifiable feet was delivered (See Figure 5).

Discussion

The TRAP sequence represents a rare but severe complication of monozygotic multiple gestations affecting 1 in 100 monozygotic twins or 1 in 35,000 pregnancies overall, and is associated with a poor prognosis.⁸ This incidence has also been found to be as high as 1 in 30 in multiple pregnancies of higher order such as in triplets.¹ A host of theories have been postulated to explain the pathogenesis of the TRAP sequence.^{9–11} However, the theory proposed by Lungu et al.¹² was favoured as it is able to link the abnormalities to a vascular disruption other than morphogenesis as postulated by other authors. According to Claudius, the development of an artery-to-artery anastomosis between the twins in the presence of a fused placenta early in the first trimester is the fundamental event in the development of the TRAP sequence. Therefore, two criteria appear to be necessary for the formation of the anastomoses and subsequent vascular disruption of one twin. The first is close proximity of the developing umbilical arteries of the two embryos on a common placenta. The second is the discordant development between these embryos leading to a reversal of arterial blood flow to the delayed twin.¹³

The impact of these vascular disruption is obvious in the recipient twin and present as loss of normally developing structures and incomplete morphogenesis of other tissues resulting in a wide range of defects such as cardiac agenesis or malformation, limb reduction, intestinal atresia and gastroschisis.¹⁴ In our case, some of these features were identified, the most striking of them are the absence of a fetal head, absence of a heart and multiple limb deformities. The pump twin is prone to congestive cardiac failure due to an increase in cardiovascular demand as a result of large acardiac mass tissue especially if it is more than 50% of the normal twin. They are prone to perinatal mortality in about 55% of cases with worst outcomes associated with maternal polyhydramnios, prematurity and an increased acardiac twin – pump twin ratio.¹⁵ This is in tandem with our case where there was polyhydramnios at presentation which was however seen to be on the decline as the pregnancy progresses. There was also a premature rupture of membrane which necessitating caesarian delivery at 30 weeks + days GA.

Acardiac twin should be considered in all monochorionic twins, abnormal fetuses with no cardiac activity, cystic hygroma, generalized oedema and malformed large discordant twin with reversed flow in the umbilical artery.¹⁶ Serial ultrasonography is done for detailed assessment of such twin pregnancies. Sherer and colleagues reported the use of the Doppler velocimetry of the umbilical cord in the

TRAP syndrome, showing a markedly abnormal peak systolic to end diastolic velocity ratio.

After exclusion of chromosomal abnormality of the pump twin, serial ultrasonography follow up is employed.¹⁷ If the estimated weight of the acardiac twin is less than one fourth of the normal twin, the prognosis is excellent without additional therapy. However, if the estimated weight of acardiac twin cannot be determined from biparietal diameter or head circumference, because of the absence of a head, a rough estimate of the weight can be determined using this formula.¹⁸

(Weight (gm) = $-1.66 \times \text{acardiac twin Length} + 1.21 \times \text{acardiac twin Length}^2$) In this index case, only the weight of the pump-twin was measured through the period of observation. However, the post-op weight of the acardiac twin was about 26% of the pump-twin (800g Vs 3000g) which according to the previous studies might be responsible for the favourable outcome.¹⁹ Some of the minimally invasive method of therapy adopted to occlude vascular supply to the acardiac twin include cord occlusion or interfetal ablation. The cord occlusion can be via embolization, cord ligation, laser coagulation, bipolar or monopolar diathermy while interfetal ablation can be performed with alcohol, monopolar diathermy and radiofrequency.²⁰

Conclusion

Conservative treatment is recommended for normal twin when acardiac twin is less than 25% of the weight of the normal twin as long as there is no sign of impending congestive heart failure. Invasive management is advised when acardiac twin is above 70% of the weight of the normal twin, under this condition the prognosis is poor. Intrafetal ablation is the best modality of treatment because it is simpler, safer and more reliable than the cord occlusion techniques.

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Conflict of Interest

No conflict of interest declared.

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