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Case Report

Twin reverse arterial perfusion sequence, a diagnostic dilemma in a low-resource setting: a case report

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ABSTRACT

Twin reverse arterial perfusion (TRAP) sequence is a unique but rare complication of monochorionic twin gestation characterized by absence of complete cardiac structure ("acardia") of one twin while the second and normal twin ("pump twin") appears normal and supplies both circulations. Diagnosis requires skill and a high index of suspicion. We report a 37-year-old gravida 3 para 2 lady who presented with an initial obstetric scan finding of twin gestation with severe polyhydramnios and single twin demise at 18-weeks gestation. Subsequent feto-maternal specialist ultrasound review at 23 weeks gestation revealed structurally normal live twin but structurally abnormal dead twin with indistinct features with possible twin-twin transfusion syndrome. Despite attempts at medical amnioreduction, she developed respiratory distress and preterm contractions following worsening polyhydramnios. She subsequently had preterm delivery at 26 weeks gestation of a live twin that suffered early neonatal death and an acardiac dead twin. This report highlights an unnecessarily missed diagnosis of a rare complication of twin gestation. It buttresses the need for training and ultrasound competence. This TRAP sequence was missed antenatally despite serial scans. Where feasible, multiple gestation requires ultrasound evaluation by subspecialty teams in feto-maternal medicine for early diagnosis to plan for adequate management. This report further highlights the need for sonographers and obstetric sonologists to deliberately evaluate for abnormalities including TRAP sequence especially in monochorionic twin pregnancies. Such evaluation will lead to early diagnosis and institution of needed management.

Keywords: TRAP sequence, Monochorionic twins, Acardiac twin, Polyhydramnios, Fetal ultrasound diagnosis

INTRODUCTION

Twin reverse arterial perfusion (TRAP) sequence is a unique but rare complication of monochorionic multiple gestation.[1,2] It was first described in the early 16th century by Benedetti (1533) and Benedictus (1539).[1,3] It is characterized by the absence of a complete cardiac structure ("acardiac") of one twin, while the second and normal twin ("pump twin") supplies both circulation.[1] It is the most severe form of twin-twin transfusion syndrome (TTTS).

Globally, the incidence is 1: 35,000 in pregnancies and 1:100 in monozygotic twin pregnancies.[1,4] Researchers, however, currently believe that with improved ultrasound techniques and the widespread assisted reproductive technology, the incidence is growing. Van Germert et al reported an incidence of about 2.6% of monozygotic twins and 1: 9/500 to 11,000 of all pregnancies.[1,5]

There are two hypotheses suggested to explain the pathogenesis of TRAP, although these are still debatable. The first is the aberrant placenta vascular pattern in the early stage of monochorionic placentation, where one foetus ("acardiac") lacking autonomous placenta blood supply is perfused by a second twin ("pump") through an artero-arterial vascular anastomosis on the placenta surface.[1,6] The blood reaches the "acardiac twin" by pressure gradient through the umbilical artery and moves away through the umbilical vein. The blood from the

"pump twin" is deoxygenated because it is reflux blood from the circulation of the "pump twin"; thus, it is a reversed arterial perfusion.[1,6] Deoxygenated blood to the " acardiac twin" results in poor morphogenesis of the " acardiac twin". Four morphologic types have been identified: acardius ancephalus (commonest type; absent head and thorax; formed abdomen and limbs); acardius amorphous (shapeless); acardius acormus (extremely rare subtypes), and acardius anceps (most morphologically developed).[1,6,7] The "pump twin" on the other hand, may suffer a bleak fate due to increased cardiac demand. Polyhydramnios, cardiac failure, and foetal demise have been widely reported as some of the complications of the "pump twin".[1,6] The second mechanism for the pathogenesis of TRAP sequence is a primary defect that may occur in cardiac embryogenesis.[1] Failure in heart formation due to chromosomal abnormality or environmental factors confers the unique support for the "acardiac twin" through anastomosis between the umbilical vessels.[1] Some investigators have identified cytogenetic discordancy in some cases of TRAP sequence, but the aetiology for such aberration is unclear.[1]

Classification of TRAP sequence was proposed by Das in the early twentieth century (1902) and is based on the morphology of the "acardiac twin".[1,6] This classification is, however, not prognostic. Wong et al then proposed a more prognostic classification based on the abdominal circumference (AC) ratio of the acardiac twin and the presence or absence of features of cardiac failure in the "pump twin".[1] The two classes proposed are: Type 1(small or medium acardiac twin; AC ratio is less than 50%) and Type 2 (large acardiac twin; AC ratio is greater than or equal to 50%).[1] These classes can be sub-grouped into subtypes a (absence of features of cardiac failure) and b (presence of features of cardiac failure), respectively.[1]

Prenatal diagnosis of TRAP sequence in modern times is seamless with the current advancement in ultrasonography. Suspicion of TRAP should be entertained when, at ultrasound examination of a monochorionic twin pregnancy is displayed and one of the fetuses has evident morphological abnormalities.[1] Pathognomonic features for TRAP sequence are the Doppler of a paradoxical circulation in the "acardiac" twin with arterial blood flowing towards, rather than away and in a caudal-to-cranial course in the abdominal aorta, which may be evident even during the first trimester.[1]

Management of TRAP sequence can be either conservative or interventional and is aimed at preserving the "pump twin", possibly to near term for delivery to assure for survival of the "pump twin".[8] Conservative management entails serial monitoring of the "pump twin" with no intervention and is reserved for type 1a TRAP sequence.[1] Type 1b may also be managed conservatively, but intervention should be promptly instituted following worsening of symptoms. Types 2a and 2b require prenatal invasive intervention. Treatment options include historical use of digoxin and ionotropic agents to control foetal cardiac failure, as well as the use of indomethacin to prevent preterm labour and reduce polyhydramnios.[1] Recent treatment options include hysterotomy to selectively deliver the acardiac twin, first reported in 1989 by Robie et al; cord occlusion through coil embolization, ligation under fetoscopy, hysterotomic incision, injection of thrombogenic agents (fibrin, glucose, embucilate gel) thermocoagulation and laser coagulation; and ultrasound guided acardiac intrafetal ablation through alcohol embolization, monopolar diathermy, laser and radiofrequency ablation.[8-10]

Recent technological advancements in therapeutic and diagnostic capabilities in maternofoetal medicine have been reported in various clinical settings, including possible prognostic factors that may influence its management, including techniques and outcomes of various modes of treatment. Although the fetal outcome of this case was poor due to low resource settings but this case highlights the need for sonographic diligent evaluation for TRAP to avert missed diagnosis. This case is being reported due to its rarity and to review the challenges in prenatal diagnosis and management in our environment.

CASE REPORT

The index case was a 37-year-old gravida 3 para 2 (with two living children) lady. She presented to our facility on self-referral following an abnormal obstetric scan finding of twin gestation with demise of one of the twins at 18 weeks of gestation.

Pregnancy was planned and achieved spontaneously. However, she had not yet registered her for pregnancy for antenatal care at any facility at the time of presentation. On self-volition, she requested an obstetric ultrasound scan earlier on the same day she presented to our facility, which revealed a viable active foetus with an estimated foetal weight of 216g at approximately 18 weeks and 4 days co-existing with a second foetus with no cardiac activity at approximately 17 weeks of gestation. Based on the result of the ultrasound scan she was counselled to present to our facility. There were no features of the TORCHES complex, and she was not a known hypertensive or diabetic. She does not smoke cigarettes nor drink alcohol. There was no attempt at termination of pregnancy, no use of herbal medications or over-the-counter drugs (OTC). She had two spontaneous vertex deliveries in 2016 and 2019, respectively of two male neonates who were alive and well.

At presentation, her physical examination findings were normal. We requested a repeat obstetric ultrasound scan, which showed twin gestation with only one viable foetus of about 18 weeks' gestation, plus polyhydramnios with an amniotic fluid index (AFI) of 30. Other routine blood and urine investigations done for her were all normal. A detailed anomaly scan also revealed the demise of the second twin, with a structural anomaly noticed in the second twin while the first twin was viable. The scan

conclusion was features suggestive of twin-twin transfusion syndrome with severe polyhydramnios.

She was counseled on the findings, and the patient opted for conservative management. She was conservatively managed through serial ultrasound monitoring and placed on tabs of indomethacin 25 mg thrice daily for three weeks as a form of medical amnio-reduction. However, seven weeks later, she presented with worsened polyhydramnios and preterm labour and subsequently had a vaginal delivery with demise of the "pump twin" (Figure 1). The placenta also showed monochorionicity (Figure 2). She was counseled on the outcome, the need for preconception care and early booking in subsequent pregnancy.

Figure 1: The photo of both twin showing TRAPS.

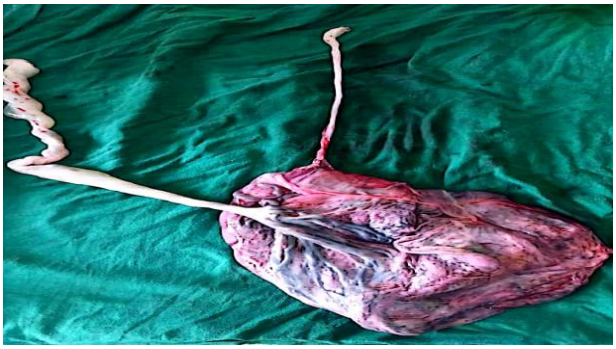

Figure 2: Showing monochorionicity.

DISCUSSION

TRAP sequence as a rare complication affecting 1-2.6% of monozygotic twin gestations and 1:9,500-11,000 of all pregnancies, is an obstetric concern that confronts fetal specialist units with varied challenges and limitations, globally.[1,4,5] However, literature is rife with successful management of the TRAP sequence in some centres with the requisite sophistication and human capacity.[5,8,9] Early ultrasonography was key in all these cases, where early detection of TRAP before 16 weeks enabled such centres to closely monitor such pregnancies and prepare for therapeutic interventions when deemed fit.[9] In our case, the patient booked late and never had antenatal care in early pregnancy, which contributed to the poor outcome.

Antenatal coverage in Africa is still at a staggering rate of 39 to 49%.[11,12] Intriguingly, this value is for the middle and high sociodemographic classes of the populace in Africa, which constitutes the higher proportion of women who seek good antenatal care.[11] Our client, unfortunately, falls outside this middle or high sociodemographic class. So, it was not surprising why she was yet to enlist her pregnancy for antenatal care in any facility at 18-weeks gestational age; more so, she had her first obstetric ultrasound scan done at same gestational age at which time the complications of TRAP was full blown with little or no chance left for successful intervention had the requisite intervention and human capacity been available. This poor seeking health (antenatal) attitude is not limited to our client but also common amongst many women in Africa especially in low income countries who for dearth of fund or knowledge resign to care at peripheral primary health centres, traditional birth attendant centres or worse still resort to homecare from supposedly experienced women resident in their locales.[11-13]

On the other hand, most of the secondary and tertiary health facilities are devoid of the practical skills and sophistication requisite to diagnose and manage some of these foetal antenatal complications found in pregnancies such as the TRAP sequence. So, had the diagnosis of TRAP sequence been made early in our client, she likely would have still suffered the same unfortunate fate simply because of the dearth of skills and sophistication to institute therapeutic intervention by most centres around her. So, her experience is that of a double-edged sword: if she had escaped the limitation of early diagnosis, she would have been stuck with the limitation of getting successful management. This is the fate of some pregnant women in low and middle-income countries in Africa.

We therefore recommend more sensitization of our eligible women to engender a good health-seeking attitude, which allows for early antenatal bookings before 12 weeks, as recommended by WHO as well as educate our peripheral centres (primary health care centres, traditional birth attendant centres) on the need for ultrasonography in pregnancy and early upscaling of care to secondary or tertiary facilities for all high-risk pregnancies.[11] Also, there should be an organized training and retraining of health care workers to build capacities requisite for diagnosis and management of fetal antenatal complications with dire perinatal morbidity and mortality.

CONCLUSION

This report describes a case twin reverse arterial perfusion sequence which was missed antenatally. She was managed conservatively and had adverse fetal outcomes. This case is being reported due to its rarity and the need for sonographers and obstetricians to deliberately evaluate for TRAPS, especially in monochorionic monoamniotic twin pregnancies, which will assist in better management of such cases.

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