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Case report-fetal major anomaly-human cyclops fetus acardia

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Abstract

Twin Reverse Arterial Perfusion Sequence (TRAP), refers to a rare, unique complication of mono-chorionic twin pregnancy in which a twin with an absent or a nonfunctioning heart (acardiac twin) is perfused by its co-twin (pump twin) via placental arterial anastomoses. The acardiac twin usually has a poorly developed heart, upper body and head. The pump twin is at risk of heart failure and problems related to preterm birth. Early diagnosis of acardiac twin by ultrasonography and color Doppler is essential for timely management and preventing complications like preterm delivery, cardiac failure and intrauterine fetal death of 'pumpco-twin'. This case report presents an antenatally diagnosed mono-chorionic twin pregnancy, resulting in preterm delivery of a healthy, normal twin along with an acardiac co-twin.

Keywords: Acardiac twin, mono-chorionic, twin reversal arterial perfusion, pump twin

Introduction

The acardiac twin or TRAP sequence is a such rare complication of monozygotic multiple pregnancy. TRAP sequence is characterized by having one normally formed "pump" twin and one acardiac (congenital absence of heart) "perfused" twin. The normal twin works as the donor and has an increased arterial perfusion pressure whereas the acardiac twin receives deoxygenated blood pumped by its co twin due to reversal of blood flow in the umbilical artery [1]. The Etiopathogenesis of acardiac anomaly is explained by the presence of an arterio-arterial anastomosis or by organ dysmorphogenesis [2] Cardiac dysmorphogenesis is developmental defect in organogenesis, a "cardiac regression sequence". The arterio-arterial anastomosis is characterized by the reversed blood flow from umbilical artery of the pump twin into the umbilical artery of the perfused twin due to sharing of a common placenta and abnormal placental vascular anastomoses between the twins [3, 4]. The pump twin is at risk of development of cardiac failure, intra uterine growth retardation, polyhydramnios or even Intra uterine death due to reversal of flow. So the aim is early diagnosis with intensive monitoring of pregnancy or interventions as and when necessary. When acardiac twin is smaller than the pump twin (< 70% of the pump twin weight) and there is no sign of cardiac decompensation in pump twin, conservative management is the best option; to prevent premature birth and to avoid complications of interventions [5].

Case report: A 28 year-old Indian woman G3P2L2 without any relevant medical history, was visited first at 12weeks of gestation for an early pregnancy ultrasound. A twin pregnancy was noted with one amorphic embryo without heart activity. The statue was diagnosed as mono chorionic di-amniotic twin pregnancy with early demise of one embryo. Follow-up was organized with routine structural ultrasound (anomaly scan) at 18 weeks of gestation, which no anomaly had been noted, but the size of the other embryo had enlarged and the cardiac activity was undetectable and no definite limbs and on follow up USG diffuse subcutaneous edema noted and liquor was nil in second baby. A reversal arterial flow was noted on a Doppler imaging study, coming from the apparently normal twin to the abnormal fetus and a diagnosis of TRAP sequence and acardiac twin made. It was decided to follow the pregnancy by further intensive monitoring of the surviving fetus and looking for eventual sign of cardiac DE compensation. The patient was admitted at 33 weeks of gestation and followed by ultrasound and two doses of 12 mg betamethasone were injected for lung maturity. Doppler investigations were normal. The biophysical profile of pump twin was reassuring.

A large (size) hypohetero-echoic mass (the amorphic fetus) without heart activity and with internal cystic calcification was seen. At 34 weeks of gestation due to rupture of membrane, termination with caesarean section was performed due to suspected large volume of the acardiac twin, primary caesarean section was planned. (Fig 1 and 2). First the surviving fetus was

delivered with breech presentation, a normal baby, weighing 1700 grams (Figure 3). After that, the acardiac twin, weighing 1800 grams, was born. The acardiac twin did not have head and no defined limbs (Figures 4-5). Examination of the placenta demonstrated vascular connections between donor and receiver (Fig 6-7).

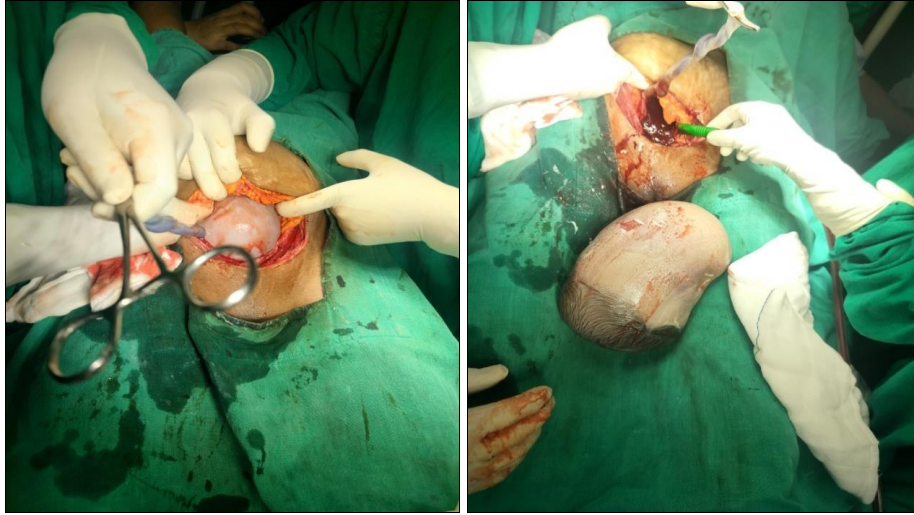


Fig 1 and 2: Cesarean section of twins with TRAP with acardia fetus



Fig 3: Normal fetus

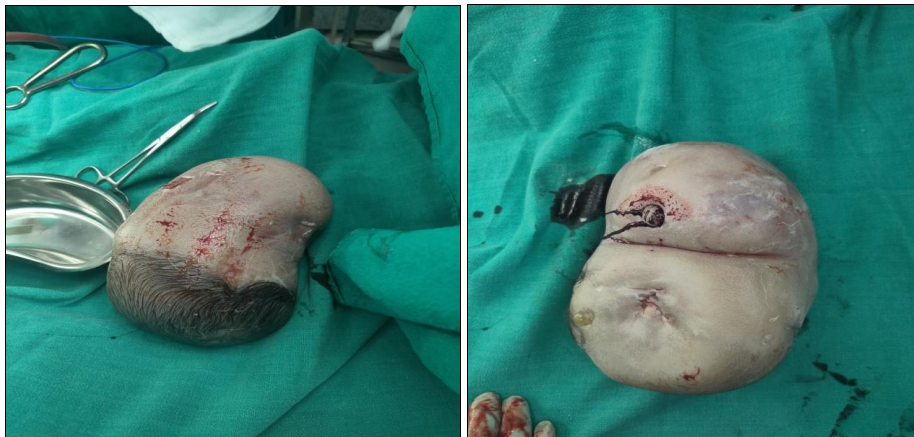


Fig 4 and 5: Acardiac twin



Fig 6: Placenta of twins with absent umbilical cord of acardiac fetus



Fig 7: X-ray of acardiac twin

Discussion

TRAP sequence was defined by Van Allen *et al.* in 1983, as a reversed perfusion from pump to acardiac twin via single anastomosis. This anastomosis can be either vein-to-vein or arterial-to-arterial, between the two cords or indirectly through the chorionic plate. Acardiac twin loses direct vascular connections with the placental villi and receives blood from pump twin through umbilical artery and the venous blood exits through umbilical vein [6].

The incidence of multi-fetal pregnancies has dramatically increased due to assisted reproductive technologies, although our case was in spontaneous conception. As a result, preterm labor, preterm premature rupture of the membrane, congenital anomalies and fetal losses are commonly encountered by obstetricians. Despite advances in diagnostic modalities and prenatal care, many times the complications associated with

multi-fetal pregnancies may remain undiagnosed. Complications are more with mono-chorionic twin gestations due to placental sharing [7].

The TRAP sequence, also known as acardiac twinning, is a malformation that occurs only in mono-chorionic pregnancies, with a frequency of about 1 per 35000 deliveries [8-10], though nowadays; the incidence appears to be higher, almost 2.6 percent of mono-chorionic twin pregnancies and 1 in 9500 to 11000 pregnancies. 4 These extremely malformed fetuses have no heart at all (holoacardia) or only rudimentary cardiac tissue (pseudoacardia) in association with multiple other developmental abnormalities [11].

Acardiac twin is classified according to the degree of morphological maldevelopment [11].

1. **Acardius-acephalus:** The acardiac twin has no cephalic development. Head and upper extremities are absent. This is

the most common type.

2. **Acardius-anceps:** The acardiac twin has cranial structures and neural tissue development. The body and upper extremities are present. This is a highly developed form of acardiac twin.
3. **Acardius-acornus:** In this, the twin has cephalic structure, but no trunk structures are present. The umbilical cord is attached to the head. This is the rarest form of the acardia.
4. **Acardius amorphous:** In this the acardiac twin has the most severe malformation with no distinguishable cephalic or truncal structure. This is least developed form and has no recognizable human structures. This differs from teratomas only by its attachment to an umbilical cord.

Conclusion

In monochorionic twin pregnancies with acardiac twin; risk of mortality of pump twin is very high. Early diagnosis, proper ultrasound surveillances for monitoring and treatment (either conservative or invasive) for the pump twin at right time is required to improve perinatal outcome. Thorough and proper monitoring, accurate diagnosis and timely interventions with co-operation and awareness from patients may help in improving the outcomes in such cases and here surviving fetus appears to be normal and recovering out from prematurity.

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Conflict of Interest: Not available

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