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Case Report

Anatomy Section

Twin Reversed Arterial Perfusion Sequence Presenting as Acardius Anceps in a Monochorionic Diamniotic Twin Pregnancy: A Case Report

MANEESHA SHARMA[1], JYOTI RANI[2], JYOTI ROHILA[3], MANPREET KAUR[4]

ABSTRACT

Twin pregnancies are associated with significantly higher risks of morbidity and mortality compared to singleton gestations. Among twin pregnancies, Monochorionic (MC) twins present a unique set of complications due to their shared placenta and vascular connections. One rare and severe complication is the Twin Reversed Arterial Perfusion (TRAP) sequence. Hereby, the authors present a case report of an acardius anceps foetus delivered by 25-year-old female with features observed on gross examination, including absence of craniofacial structures, an incomplete calvarium, a hypoplastic thorax lacking the heart and lungs and syndactylous limbs, while the gastrointestinal tract was preserved. The MC diamniotic placenta showed two umbilical cords on a shared chorionic plate. Histology demonstrated mature tertiary villi with syncytial knots, intact trophoblastic layers, decidual tissue and focal villous calcification. The present case underscores the severity of the TRAP sequence and the critical importance of early prenatal diagnosis and intervention. Delayed recognition, as seen in present case, can result in fatal consequences for the pump twin.

Keywords: Decidual tissue, Perinatal mortality, Placental anastomosis, Pump twin, Syncytial knots

CASE REPORT

A 25-year-old Gravida 5, Para 1. (G5P1) female with three prior spontaneous abortions and one previous normal vaginal delivery was admitted at 38 weeks' gestation with no prior history of hypertension or diabetes mellitus. The foetal anomaly scan (also called Level II ultrasound), typically performed between 18-22 weeks of gestation to detect gross congenital malformations, was not conducted during this pregnancy. The scan done at 38 weeks of gestation revealed one viable foetus with a single loop of the umbilical cord around the neck and a second non viable foetus corresponding to an estimated gestational age of 23-24 weeks. After two days of admission in the hospital, caesarean delivery was performed. Following delivery, the non viable foetus weighed around 1.5 kg and the viable twin weighed 3 kg. The viable twin initially cried but exhibited rapid clinical deterioration over the next 48-72 hours. Echocardiography revealed right ventricular hypertrophy, patent ductus arteriosus with a left-to-right shunt and dilated right atrium and ventricle, indicative of congestive heart failure. Despite medical management, the neonate succumbed to heart failure seven days after delivery.

The acardiac twin exhibited the following anomalies on gross examination: a poorly developed head with hair, an incomplete calvarium, absence of facial structures, a shallow thoracic cavity with absence of the heart and lungs and poorly developed limbs with syndactyly. The gastrointestinal tract and its associated structures were well-formed, as seen in gross examination [Table/Fig-1].

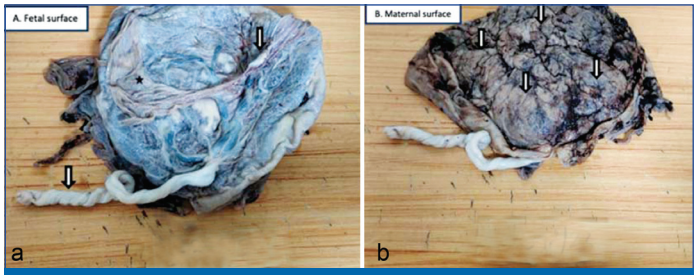
Four acardiac twin variants are classified according to the extent of cranial and torso development [1]:

1. Acardius anceps: Partial head and facial structures present.
2. Acardius acephalus: Complete absence of cranial features.
3. Acardius acormus: Head present without trunk (rarest form).
4. Acardius amorphous: Amorphous mass with mixed tissues but no distinct organs.

[Table/Fig-1]: Gross pathological findings as seen on autopsy: Presence of a poorly developed head with hairs, incomplete calvarium, absence of facial structures, shallow thoracic cavity with absence of heart and lungs, poorly developed limbs with syndactyly while Gastrointestinal (GIT) was well-formed.

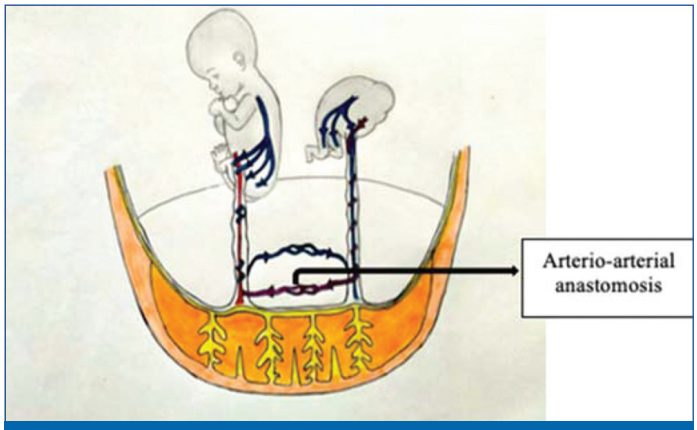
The features in present report are consistent with the acardius anceps subtype-characterised by partial cranial development and rudimentary limbs.

Gross examination of the placenta revealed a MC diamniotic configuration, consistent with a twin gestation. The foetal surface showed two distinct umbilical cords inserted into a single placental mass, confirming a shared chorionic plate [Table/Fig-2a]. The maternal surface appeared dull, with intact cotyledons, suggesting preserved placental architecture without evidence of infarction or abruption [Table/Fig-2b].



[Table/Fig-2]: Monochorionic diamniotic placenta: a) Foetal surface shows two attached umbilical cords (arrows) and membrane (star); b) Maternal surface is dull with intact cotyledons (arrows).

Of clinical significance, one of the twins was identified as an acardiac twin, lacking a functional heart and was perfused retrograde by the co-twin (referred to as the pump twin) through an arterio-arterial anastomosis, consistent with TRAP sequence. This vascular anomaly reflects abnormal hemodynamic circulation within the shared placenta and underpins the pathogenesis of the acardiac twin [Table/Fig-3].



[Table/Fig-3]: Pump twin and acardiac twin with Arterioarterial (A-A) anastomosis.

Histological evaluation of Haematoxylin and Eosin (H&E) stained placental sections (original magnification ×200) demonstrated:

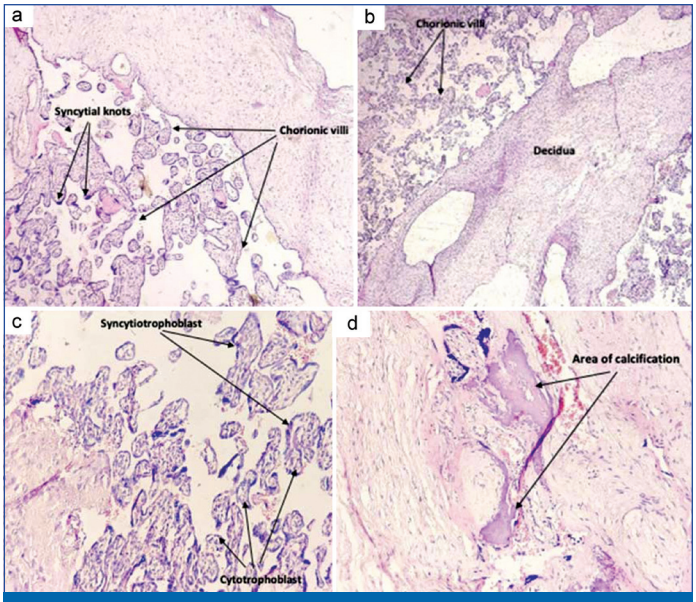
Numerous tertiary chorionic villi, many showing prominent syncytial knots, indicative of advanced villous maturation [Table/Fig-4a]; Decidual tissue adjacent to chorionic villi, maintaining a normal maternal-foetal interface [Table/Fig-4b]; Syncytiotrophoblasts and cytotrophoblasts are singular, remove 's' from the end as written below- Chorionic villi with intact dual trophoblastic layers- outer syncytiotrophoblast and inner cytotrophoblast [Table/Fig-4c]; and Focal calcifications within the villous stroma, suggestive of physiological maturation or early senescence [Table/Fig-4d].

These findings are consistent with a MC diamniotic placenta complicated by TRAP sequence.

DISCUSSION

Twin gestations confer substantially elevated perinatal morbidity and mortality compared to singleton pregnancies [1]. The TRAP sequence constitutes a rare, life-threatening complication of MC twin pregnancies, with an estimated incidence of 1:35,000 overall [1].

The pathogenesis of TRAP sequence is believed to involve abnormal embryogenesis associated with vascular anastomoses that develop within the shared placenta during early foetal development. In affected MC pregnancies, artery-to-artery and vein-to-vein placental anastomoses establish a circulatory connection between the pump twin and the acardiac twin. Through these vascular communications, the acardiac twin receives poorly oxygenated blood from the pump twin via reversed arterial flow [1].



[Table/Fig-4]: Microscopy of the placenta: a) Microphotograph shows numerous tertiary chorionic villi with syncytial knots (H&E; 200X); b) Microphotograph shows decidual tissue along with chorionic villi (H&E; 200x); c) The chorionic villi show external syncytio-trophoblast lining and an internal cytotrophoblast lining (H&E; 200X): d) Area of calcification also noted (H&E; 200X).

Batoy A et al., reported monochorionic diamniotic TRAP with an acardiac, acerebral twin and rudimentary limbs and a surviving pump twin to term delivery [2]. Thirupathi K and Nathan JM described Monochorionic Diamniotic acephalus with intestinal structures and pump twin Neonatal Intensive Care Unit (NICU) survival despite cardiomegaly [3]. Nicoli P et al., presented two Monochorionic Monoamniotic (MCMA) TRAP sequence cases, both featuring acephalic acardiac twins. One of the cases revealed an acephalic acardiac twin presenting as an amorphous mass with rudimentary lower limbs and feet, generalised oedema and confirmed placental arterio-arterial anastomoses. No cardiac or cranial structures identified. Another case demonstrated absent cardiac activity and reversed flow in the acardiac twin; Histopathological examination confirmed MCMA placentation with vascular anastomoses diagnostic of TRAP [4]. Sayanthan B et al., reported a parabiotic acardiac twin in TRAP sequence, emphasizing vascular interdependence and poor pump prognosis without intervention [5]. Unlike these, the present case uniquely preserved enteric structures amid profound craniothoracic defects.

Management of TRAP sequence is guided by the size of the acardiac twin and the clinical status of the pump twin. Expectant management with close ultrasonographic surveillance may be appropriate when the acardiac twin is small and the pump twin shows no evidence of haemodynamic compromise. In contrast, foetal intervention, including radiofrequency ablation, laser coagulation, or bipolar cord occlusion, is indicated when the acardiac twin exceeds 70% of the pump twin's estimated weight, or when signs of cardiac overload, progressive polyhydramnios, hydrops fetalis, or rapid growth of the acardiac twin are present. Prognosis is primarily influenced by early diagnosis, the acardiac-to-pump twin size ratio and the cardiovascular condition of the pump twin, with timely intervention significantly improving survival outcomes [5].

CONCLUSION(S)

Twin pregnancies, particularly monochorionic gestations, carry an elevated risk of complications due to shared placental vasculature. This case underscores the severity of the TRAP sequence and the critical importance of early prenatal diagnosis and intervention. Delayed recognition, as seen in present case, can result in fatal consequences for the pump twin. Continued documentation and

anatomical study of such cases are vital for advancing clinical understanding and guiding future management strategies.

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PARTICULARS OF CONTRIBUTORS:

1. Professor and Head, Department of Anatomy, AIMS, Mohali, Chandigarh, Punjab, India.
2. Demonstrator, Department of Anatomy, MMIMSR, Mullana, Ambala, Haryana, India.
3. Assistant Professor, Department of Anatomy, AIMS, Mohali, Chandigarh, Punjab, India.
4. Senior Resident, Department of Pathology, AIMS, Mohali, Mohali, Punjab, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:
Jyoti Rohila,
Assistant Professor, Department of Anatomy, Dr. B. R. Ambedkar State Institute of Medical Sciences (AIMS), Mohali (S.A.S. Nagar)-160055, Punjab, India.
E-mail: jyotirohilla5671@gmail.com

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