Taiwanese Journal of Obstetrics & Gynecology 60 (2021) 177–180

Contents lists available at ScienceDirect

Taiwanese Journal of Obstetrics & Gynecology

journal homepage: www.tjog-online.com

Case Report

Twin reversed arterial perfusion syndrome in a monochorionic monoamniotic twin pregnancy

Yi-Yan Chen [a, 1], Chien-Chu Huang [a, b, 1], Chih-Yi Yang [a, c], Tsan-Hung Chiu [a, d], Ming Ho [a, d, *]

[a] Department of Obstetrics and Gynecology, China Medical University Hospital, No. 2, Yuh-Der Road, Taichung, 404, Taiwan
[b] Graduate Institution of Biomeidcal Sciences, China Medical University, No. 2, Yuh-Der Road, Taichung, 404, Taiwan
[c] Department of Public Health, China Medical University, No. 2, Yuh-Der Road, Taichung, 404, Taiwan
[d] Department of Medicine, China Medical University, No. 2, Yuh-Der Road, Taichung, 404, Taiwan

ARTICLE INFO

Article history:
Accepted 13 April 2020

Keywords:
TRAP
Twin-Reversed Arterial Perfusion sequence
Acardiac twin
Monochorionic twin

ABSTRACT

Objective: Twin-Reversed Arterial Perfusion (TRAP) sequence is a rare complication of monochorionic multiple gestation. Conservative management should be considered if there is no poor prognostic factor.
Case report: This is a 35 year-old female with twin pregnancy with acardiac monster. Under the request of the patient, there was no intervention during the whole pregnancy. We keep regular and close sonography weekly follow up. There was no maternal complication and there was also no heart failure sign or polyhydramnios of the donor twin. Minimal blood flow was noted at the anastomotic vessels under the sonography at late gestational age. Due to breech presentation, cesarean section was performed at gestational age 37 + 1/7 weeks. She delivers a healthy baby smoothly.
Conclusion: Antenatal sonography is an important tool to evaluate the fetus status. Under special condition, term pregnancy is still possible without any treatment.
Case report: Twin reversed arterial perfusion syndrome in a monochorionic monoamniotic twin pregnancy.

Introduction

Acardiac twinning, or the twin reversed arterial perfusion (TRAP) sequence, was first reported by Doctor Beneditti in 1533. It occurs in about 0.3–1% of monozygotic twins, and about 1 in 35,000 births. The risk of recurrence is estimated 1:10000. [1-3].

Pathophysiologically, it is due to the development of arterio-arterial vascular anastomoses between the umbilical arteries of monozygotic twins early in embryogenesis that cause this special condition [1]. In TRAP sequence, blood goes into the acardiac twin via the umbilical artery and leaves the acardiac twin via the umbilical vein. The blood flow directions are totally opposite form the normal physiological placenta-fetal circulation. And that's why we called it; twin reversed arterial perfusion (TRAP) sequence. Due to one twin lack of a well-formed cardiac structure, it is like a parasite and totally dependent on the other twin for entire blood supply. And the acardiac twin loses direct vascular connection with the placental villi.

Without early detection, close follow-up, and on-time intervention, mortality rates for the donor twins is up to 50–70% [2].

We report a case of a primigravida, who transfer to our hospital at 23 weeks gestational age. Ultrasound imaging showed twin pregnancy with one fetus with low extremities only.

Case

This 35 year-old female with gravida 1 parade 0 abortion 0 was referred to our hospital due to twin pregnancy at about 23 weeks gestational age with one fetus with low extremities only. There was no history of alcohol drinking, drug abuse, smoking, or infection during this pregnancy. She was a housewife with average height, weight. And she had no previous medical or cousin marriage history.

* Corresponding author. Department of Obstetrics and Gynecology, China Medical University Hospital, No. 2, Yuh-Der Road, Taichung, 404, Taiwan.
E-mail addresses: d20092@mail.cmuh.org.tw (Y.-Y. Chen), akamarucoh1116@gmail.com (C.-C. Huang), d24455@mail.cmuh.org.tw (C.-Y. Yang), d3861@mail.cmuh.org.tw (T.-H. Chiu), d20267@mail.cmuh.org.tw (M. Ho).
[1] Yi-Yan Chen and Chien-Chu Huang are co-first authors.

https://doi.org/10.1016/j.tjog.2020.11.029

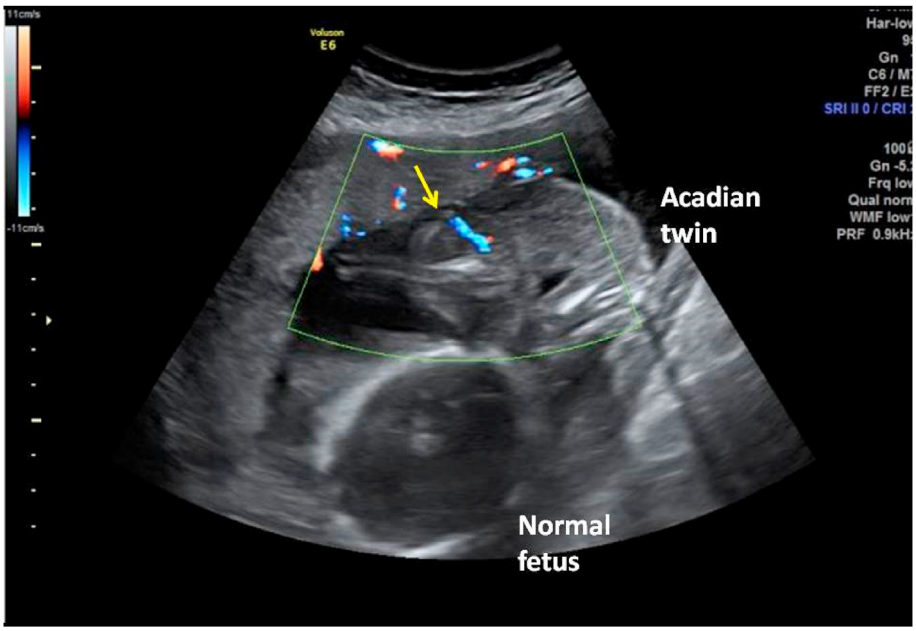


Fig. 1. Ultrasonography showing malformations in the twin fetus (Arrow points to the malformation connected vessels, consistent with the figure 3-1 grossly picture).

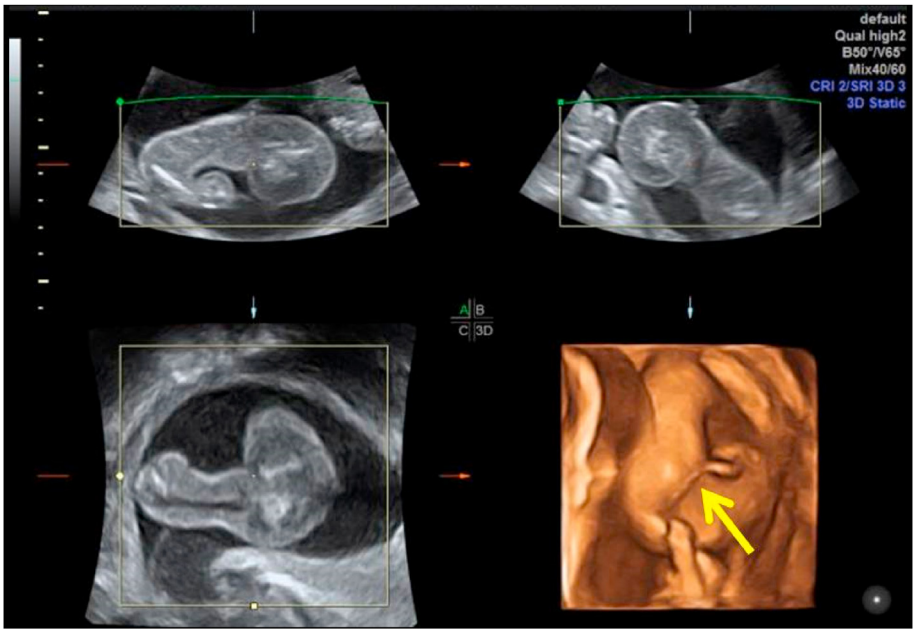
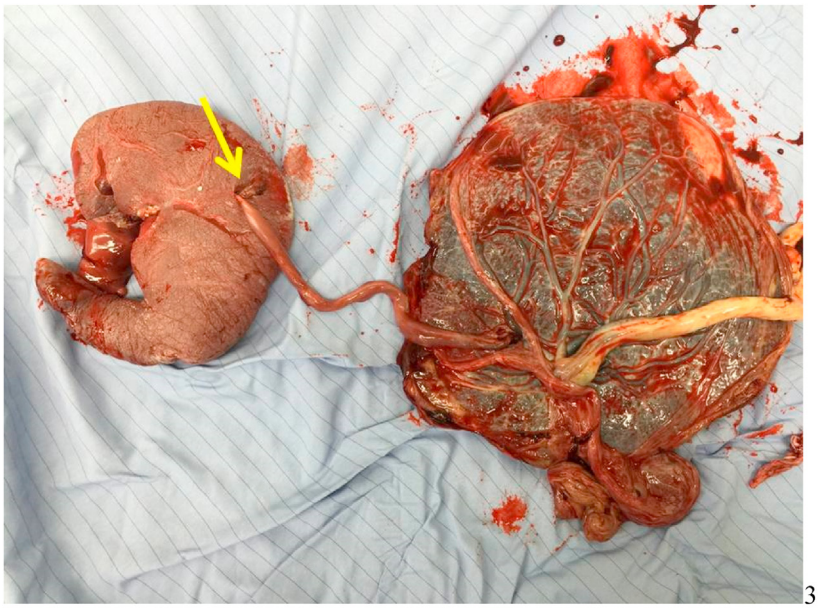


Fig. 2. A hypo-hetero-echoic mass with femoral bone (the acardiac monster) without heart activity in ultrasound. (Arrow points to the malformation connected vessels linked to the acardiac monster).

This time, she got twin pregnancy naturally. However, about 23 week's gestational age, twin with one fetus with low extremities only was noted. She then transferred from local clinic to our outpatient department. Investigations with complete blood count, urinalysis, and blood glucose measurement was done and the result was within acceptable range. Ultrasonography revealed a monozygotic twin pregnancy with one acardiac fetus and one normal fetus with no visible malformation (BPD = 5.29 cm HC = 20.64 cm AC = 17.97 cm FL = 3.67 cm AFI = 11.79 cm). Acardiac twin with lower limbs only and the other twin had no congestive heart failure or polyhydramnios. Estimated fetal weight was 500 g for the pump twin. Ultrasonography finding was consistent with twin reversed arterial perfusion (TRAP) sequence. TTTS with acardiac monster was diagnosed (Figs. 1,2). Intra-fetal ablation was suggested. But the patient decided to do observation first.

The pregnancy course was smoothly. We keep regular follow up for the fetus status. Rechecked ultrasonography at 31 + 4/7 weeks gestational age revealed acardiac fetus (FL = 4.60 cm) and one normal fetus without congestive heart failure or polyhydramnios (BPD = 7.85 cm HC = 29.38 cm AC = 24.71 cm FL = 5.55 cm AF largest pocket = 3.19 cm). Delivery was suggested between 34 and 36 weeks gestational age, but the patient want to delivery after 37 weeks gestational age. In the whole course, there was no headache, nausea, vomiting, blurred vision or limbs edema noted. She was admitted at 37 + 1/7 weeks gestational age for delivery.We arranges ultrasonography every week. Minimal blood flow was noted at the


3-1


3-2

Fig. 3. Acardiac monster, (3-1) placenta (Arrow points to the malformation connected vessels, consistent with the Fig. 1 sonography picture) and(3-2) the donor twin fetus at birth.

anastomotic vessels under the sonography at late gestational age. Due to twin pregnancy with reversed arterial perfusion (acardiac monster) and breech presentation, lower segment transverse cesarean section was performed under spinal anesthesia. Twin A, female baby was delivered smoothly with pediatrician standby. Her weight was 2735 gm and the apgar score was 7′ to 8'. The Placenta weight 610 gm with normal appearance, MCDA type. Examination of the placenta showed vascular connections between donor and receiver. Twin B was acardiac monster with lower limbs only (Fig. 3).

Discussion

About 75% of monozygotic twins share the same placenta and the anastomoses of the placental can cause serious complications. Acardiac twins, or the twin reversed arterial perfusion (TRAP) sequence is a rare but serious one. There are four kinds of acardiac twins including: acardiac anceps, acephalic acardiac fetus, acardiac acormus, and acardiac amorphus. The donor twin may die of prematurity and respiratory distress syndrome (RDS) after birth. The overall mortality of donor twin is approximately 50–75%. But the accurate risk prediction is currently hard to calculate.

When to start therapeutic treatment in TRAP-sequence is still controversial [3].Observation of the pregnancy under weekly ultrasounds and the intrauterine interventions can increase the survival chances of the donor twin. One study also suggested pump/acardiac umbilical venous diameter (UVD) ratios, probably could allow risk prediction of pump twins [4].

However, without proper management, the mortality rate of TRAP sequence is ranging from 35% to 55%. Once invasive treatment is indicated, the safe and effect treatment should be offer as soon as possible [1]. Earlier intervention and even prophylactic intervention in the first trimester have been discussed. The interventions for TRAP sequence include amniodrainage for the polyhydramnios, and aggressive treatment of the acardiac twin like hysterotomy with selective delivery, intrafetal ablation, and funicular occlusion [5]. Intrafetal ablation can be applied with alcohol, laser, thermocoagulation or radiofrequency. Funicular occlusion can be done under ultrasound-guided or fetoscope-guided. The methods of funicular occlusion include ligation, laser, intrafunicular embolization with foreign bodies and thermocoagulation. One study also shows that high-intensity focused ultrasound (HIFU) therapy is beneficial and can reduce the cardiac load of the pump fetus [6]. All the method is to cut the blood connection between the twins and protect the donor twin. By using minimally invasive treatment, ligation for the vascular anastomosis at early gestational weeks reveals a benefit in improving the outcome of the donor twin.

However, not all the TRAP sequence needs treatment. When there is no poor prognostic sign, like heart failure sign and polyhydramnios, conservative treatment can be considered. Recent studies reveal that spontaneous cessation of blood connection does happen, and the acardiac twin will be smaller than the donor twin [7]. Under this special condition, good outcome of the donor twin is noted [8].

In our case, the patient and family refused interventions, and expectant management was done under fully explanations of the every possible condition and poor outcome. We performed weekly Color-Doppler sonography examination for monitoring the fetus status.There was no complication of the donor twin and the deliver was arranged at term. Minimal blood flow was noted at the anastomotic vessels at late gestational age may play a key role. How does spontaneous cessation of blood connection happen is unknown. There is some case reports showed the similar process but not till term delivery. Our case shows that under conservative follow up only, the TRAP sequence can be alive without complication till term birth. Although the risk of complications is high, some select cases still can be managed conservatively.

Funding

There is no funding or support in this study.

Declaration of competing interest

The authors whose names are listed immediately below certify that they have NO affiliations with or involvement in any organization or entity with any financial interest (such as honoraria; educational grants; participation in speakers' bureaus; membership, employment, consultancies, stock ownership, or other equity interest; and expert testimony or patent-licensing arrangements), or non-financial interest (such as personal or professional relationships, affiliations, knowledge or beliefs) in the subject matter or materials discussed in this manuscript.

Acknowledgments

We thank our colleagues from department of Obstetrics and Gynecology, China Medical University Hospital.

References

[1] Chen C-P. Acardiac Twinning (Twin reversed arterial perfusion sequence): a review of prenatal management. Taiwan J Obstet Gynecol 2005;44(2):105–15.
[2] James WH. A note on the epidemiology of acardiac monsters. Teratology 1977;16(2):211–6.
[3] Mone F, Devaseelan P, Ong S. Intervention versus a conservative approach in the management of TRAP sequence: a systematic review. J Perinat Med 2016;44(6):619–29.
[4] van Gemert MJ, Pistorius LR, Benirschke K, Bonsel GJ, Vandenbussche FP, Paarlberg KM, et al. Hypothesis acardiac twin pregnancies: pathophysiology-based hypotheses suggest risk prediction by pump/acardiac umbilical venous diameter ratios. Birth Defects Res A Clin Mol Teratol 2016;106(2):114–21.
[5] Tan TY, Sepulveda W. Acardiac twin: a systematic review of minimally invasive treatment modalities. Ultrasound Obstet Gynecol 2003;22(4):409–19.
[6] Seo K, Ichizuka K, Okai T, Dohi S, Nakamura M, Hasegawa J, et al. Twin-reversed arterial perfusion sequence using high-intensity focused ultrasound therapy. Ultrasound Obstet Gynecol; 2019 Jul;54(1): p. 128–34.
[7] Sepulveda W, Sebire N. Acardiac twin: too many invasive treatment options—the problem and not the solution. Ultrasound Obstet Gynecol 2004;24(4):387–9.
[8] Dubey S, Verma M, Goel P, Punia R, et al. Twin reversed arterial perfusion: to treat or not? J Clin Diagn Res 2017;11(1):QD05–7. https://doi.org/10.7860/JCDR/2017/24400.9140.