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LATION PERFORMED ON AN ACARDIAC TWIN WITH TRAP SEQUENCE: A CASE REPORT

LASER PHOTOCOAGU-

Coridon Franco da Costa

Professor of Gynecology and Obstetrics; Gynecologist and Obstetrician at Hospital Santa Casa de Misericórdia de Vitória/ES, Brazil

Antônio Chambô Filho

Professor of Gynecology and Obstetrics; responsible for the Gynecology and Obstetrics Service at Hospital Santa Casa de Misericórdia de Vitória/ES, Brazil

Barbara Eugenio Custodio Silva

Resident Doctor of the Gynecology and Obstetrics Service of the Santa Casa de Misericórdia Hospital in Vitória/ES, Brazil

Heloisa Rocha Ribeiro

Resident Doctor of the Gynecology and Obstetrics Service of the Santa Casa de Misericórdia Hospital in Vitória/ES, Brazil

Isadora Favarato Sagrillo

Resident Doctor of the Gynecology and Obstetrics Service of the Santa Casa de Misericórdia Hospital in Vitória/ES, Brazil

Catarina Epichin Amin

Medical Student at Escola Superior de Ciências da Santa Casa de Misericórdia de Vitória (EMESCAM). Vitória/ ES, Brazil

Elisa Smith Barbiero Medeiros

Medical Student at Escola Superior de Ciências da Santa Casa de Misericórdia de Vitória (EMESCAM). Vitória/ ES, Brazil



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Iúlia Cera Scotá Moreira

Medical Student at Escola Superior de Ciências da Santa Casa de Misericórdia de Vitória (EMESCAM). Vitória/ ES, Brazil

Luise Schwan Soares

Medical Student at Escola Superior de Ciências da Santa Casa de Misericórdia de Vitória (EMESCAM). Vitória/ ES, Brazil

Abstract: Introduction: Monochorionic twin pregnancies complicated by reversed arterial perfusion (TRAP sequence), in which a "pump" twin perfuses an acardiac twin through aberrant arterio-arterial anastomoses, require intervention considering the risk for the "pump" twin. Given the scarcity of reports on this pathology, this article aims to report the case of laser photocoagulation performed on an acardiac twin with TRAP sequence and its outcomes until delivery. Case presentation: 34 years-old, woman, healthy, with positive family history of twinning and obstetric history of 4 pregnancies, 3 previous normal births without complications and no miscarriages. The ultrasound at 15 weeks and 5 days showed findings of a morphologically normal fetus and another acardiac. On spectral Doppler of the abdominal insertion of the umbilical cord of the acardiac fetus, reverse flow was visualized, in addition to increased resistance to flow in the venous duct of the "pump" fetus, confirming the diagnosis of the TRAP sequence. At 19 weeks and 6 days of gestational age, laser photocoagulation of the umbilical cord of the acardiac twin was performed via transamniotic surgery guided by ultrasound, using a laser fiber passed through a needle that sustained interruption of the vascular flow. The pregnancy continued without complications and at 37 weeks and 0 days, it was interrupted by cesarean section, as recommended by the medical team. Conclusion: The patient progressed well after the procedure and monitored with weekly ultrasounds that showed a reduction in reverse blood flow and the survival of the pump twin. The cesarean section was performed without complications. In terms of survival of the pump fetus and reduction of complications, early intervention is suggested. Also, the main intervention methods are not superior when compared to each other and the center's resources, clinical presentation, gestational age, operator experience and team preferences should be considered when choosing the method.

Keywords: Acardiac twin; Laser Coagulation; Pregnancy, Twin; TRAP sequence.

INTRODUCTION

In monochorionic twin pregnancies, a rare complication known as twin reversed arterial perfusion sequence (TRAP) can occur, in which a co-twin ("pump" twin) perfuses an anomalous twin with an absent heart (acardiac twin) through aberrant arterio-arterial anastomoses.1 The acardiac twin depends on the circulatory support provided by the pump twin, and the upper body and head are generally underdeveloped or even absent,1 with no potential for survival outside the womb. By perfusing the acardiac twin, the pump twin can develop anemia, heart failure, and fetal hydrops.² In addition, in the absence of intervention, premature birth or intrauterine death can occur.1

Currently, many countries perform successful interventions in the amniotic cavity to treat TRAP complications, saving the lives of 80% of pump twins.2 Since the acardiac twin has no significant potential for long-term survival, treatment of the TRAP sequence focuses entirely on the pump twin.1 Cord occlusion therapy for managing the TRAP sequence is designed to interrupt vascular communication between the twins, thereby reducing the hemodynamic burden of the pump twin and obstructing blood flow to the acardiac twin.1 Intervention leads to a better prognosis than conservative treatment, especially in pregnancies with one or more unfavorable characteristics.1

Given the scarcity of reports on this pathology and the possibility of intrauterine treatment, this article aims to report the case of laser photocoagulation performed on an acardiac twin with TRAP sequence detected and its outcomes until delivery.

CASE PRESENTATION

A 34-year-old woman, without any known comorbidities or addictions aside from anxiety and continuous use of sertraline, with a positive family history of twinning, blood group A positive. She had an obstetric history of four pregnancies, three previous normal births 14, 10, and 5 years ago, without complications, and no miscarriages.

The first transvaginal obstetric ultrasound scan showed a single, topical gestational sac with embryo A: fetal heartbeat (FHR) of 135 bpm and crown-rump length (CRL) of 4.4 mm; and embryo B: no FHR and CRL of 3.7 mm, compatible with a monochorionic and diamniotic pregnancy estimated at six weeks and one day of gestation.

The 1st-trimester morphological ultrasound carried out at 11 weeks and five days showed fetus A to be morphologically normal, with body movement and nasal bone present, CRL of 57.7 mm, FHR 146, and nuchal translucency (NT) of 1.9 mm, and fetus B with multiple structural malformations, absence of heart and pronounced edema (fetus B: acardiac fetus). Double amniotic cavity, normal volume, single placenta, posterior, grade 0. Monochorionic and diamniotic twin pregnancy, with fetal biometry at 12 weeks and two days and the presence of TRAP. In addition, the doppler of the ductus venosus of fetus A had a normal pulsatility index (PI) (0.74), and the doppler of the uterine arteries also showed normal flow (mean PI of 1.72).

After the diagnosis of the TRAP sequence, the patient was referred to the fetal medicine outpatient clinic for evaluation, where she underwent a new ultrasound at 15 weeks and five days. This scan showed morphologically normal fetus A (Figure 1), and spectral Doppler of the abdominal insertion of the umbilical cord of the acardiac fetus (Figure 2) showed reverse umbilical cord flow (Figure 3), as well as increased resistance to flow in the venous duct of the "pump" fetus, once again corroborating the diagnosis of TRAP sequence.



Figure 1: Three-dimensional ultrasound showing "pump" fetus and acardiac fetus.



Figure 2: Insertion of the umbilical cord of the "pump" fetus (A) and insertion of the abdominal umbilical cord of the acardiac fetus (B).

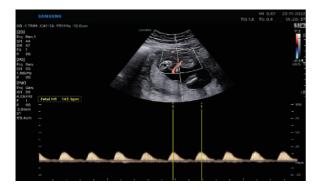


Figure 3: Color and spectral Doppler of the umbilical artery (acardiac twin), showing reverse blood flow.

At a follow-up ultrasound appointment, it was found that the proportion of the weight of the acardiac fetus compared to the weight of the pump fetus exceeded 0.5. Therefore, the attending physician indicated interven-

tion and advised the patient about the risks and benefits of the procedure to be conducted and consulted about consent. Thus, at 19 weeks and six days of gestational age, laser photocoagulation was performed in the maternity ward on the abdominal insertion of the umbilical cord of the acardiac twin via the transamniotic route. The procedure was conducted with ultrasound guidance, using a 17-gauge spinal needle guided by hand, directed at an intrafetal tissue target adjacent to the umbilical cord vessels (Figure 4). A laser fiber was passed through the needle and, over a series of short controlled bursts, sustained interruption of vascular flow was achieved. The equipment used was the INNOVA Touch Duo model (Orligth laser). The procedure was conducted under spinal anesthesia without complications and was considered successful after confirmation of the absence of umbilical cord flow at the ablation site. Cephalexin 250 mg was administered orally approximately 1 hour before the procedure and continued every 6 hours for 72 hours.

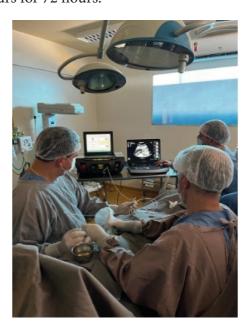


Figure 4: Laser photocoagulation procedure in an acardiac twin with TRAP sequence.

The patient had a good post-operative evolution and was re-evaluated with an ultrasound six days after the procedure, with an estimated gestational age of 20 weeks and five days. This ultrasound study showed a "pump" fetus with fetal movement present, FHR 152 bpm, with a 12 mm pyelocaliceal dilation on the right, with adequate amniotic fluid volume for gestational age. The acardiac fetus had a volume of 137 cm³, with flow still evident, but less when compared to previous exams, weighing 32% of the "pump" fetus.

Ultrasound monitoring occurred weekly, with gestational monitoring as part of the prenatal routine. At 33 weeks, the prenatal doctor advised the patient to undergo antenatal corticosteroid therapy with betamethasone 12 mg, intramuscularly, in two doses with a 24-hour interval between each, to achieve fetal maturity, given the risk of premature labor and the possibility of terminating the pregnancy before term due to decompensation of the "pump" fetus. However, continuous monitoring showed no abnormalities. In total, 11 prenatal consultations were conducted with no other alterations or complaints and negative serologies.

The pregnancy continued uneventfully until 36 weeks and six days, with a reduction in reverse blood flow. The newborn was delivered by cesarean section based on the recommendation of the medical team at 37 weeks. The live newborn ("pump" twin), female, was born weighing 2,745 grams, APGAR score 08/08 (5'/10'), with a length of 44 cm and cephalic, thoracic, and abdominal perimeters measuring 34 cm, 29.5 cm, and 31 cm, respectively. Physical examination revealed a frontal prominence and three preauricular appendages on the left. The acardiac twin, with no signs of life, presented macroscopically: rudimentary cephalic pole, trunk, and lower pole, weight of 1,955 grams, 71% "pump" fetus (Figure 5 A and B).

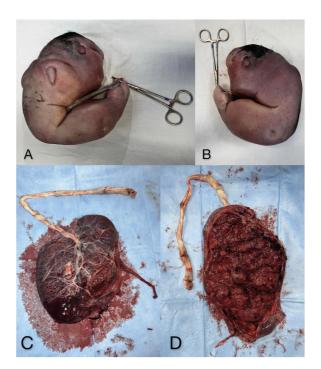


Figure 5: Macroscopic appearance of an acardiac twin and the placenta.

DISCUSSION

Fetal acardia syndrome with TRAP sequence is a rare obstetric condition with a variable incidence, although increasingly frequent - both due to the growing number of twin pregnancies (mainly by artificial fertilization methods) and due to more accessible and accurate diagnostic methods.^{1, 3, 4} More recent studies suggest a prevalence of around 1: 9,500 - 11,000 in twin pregnancies in general and 1 - 3% of all monochorionic pregnancies.^{5, 6, 7}

With advances in ultrasound technology, diagnosis is done earlier and earlier, increasing the chances of a beneficial and successful intervention.² To define the most appropriate course of action, the attending doctor should actively research some differential diagnoses. Intrauterine death of fetuses with extreme malformations is one of them, characterized by the absence of growth on ultrasound follow-up.^{4, 5} Other rarer options would be placental or intra-amniotic tumors, such as chorioangioma, which have low-resistance

pulsatile flow, or placental teratomas, which generally have no umbilical cord connection.⁴

Regarding the TRAP sequence, following ultrasound findings are considered pathognomonic: 1) the presence of an acardiac fetus perfused by an anatomically normal fetus, with 2) retrograde perfusion flow in the umbilical cord and descending aorta, through 3) the presence of arterio-arterial anastomoses. 2, 5, 8, 9, 10 Considering the pregnancy reported, the condition was initially suspected at six weeks and one day, when an acardiac fetus was identified, and was diagnosed on ultrasound at 11 weeks and five days. The patient was then referred to the fetal medicine reference center at 15 weeks and five days, when it was confirmed by the presence of the three points described above.

Concerning the morphology of the acardiac fetus, the main classification used categorizes four distinct groups,11 as follows: (a) Acephalus: absence of cephalic structures, with the presence of lower limbs, the most frequent presentation in 60-75% of cases;8 (b) Anceps: more developed form with formed body and extremities, but skull and face have deformities; (c) Amorphous: less differentiated form, mass with indistinguishable structures; (d) Acormus: only the cephalic structure is distinguishable, being the rarest group with around 5% of acormus.^{6, 8} It should be noted that these classifications have no prognostic value or predict management options.4 In the case reported, we consider that the classification that best fits morphologically is that of amorphous fetus due to the rudimentary structures that are difficult to identify in terms of limbs and cranial structure.

Evaluating the acardiac fetus can be difficult depending on its morphology, but analyzing certain parameters in both fetuses can represent prognostic indicators and help guide management. The size of the acardiac twin plays a significant role in possible complications that threaten the pump fetus, especially high-output heart failure.4, 12 In this sense, assessing the cardiovascular condition of the pump fetus suggests a worse prognosis when polyhydramnios, cardiomegaly, pericardial and pleural effusions, ascites, and tricuspid regurgitation are present.^{12, 13} Concerning the weight ratio between acardiac and pump fetuses, the risk increases when > 0.7.12 Some studies also suggest using the ratio between abdominal circumferences, considering large acardiac fetuses (≥50%) as a worse prognosis for the pump fetus, requiring interventions regardless of cardiovascular alterations.5, 13 The procedure in question was conducted from the cut-off point of 0.5 (50%) of the weight ratio between the pump and the cardiac fetus, in line with international guidelines.⁵

In Brazil, fetuses with signs indicating a poor prognosis, with a weight discordance > 0.7 and/or hydrops, are considered candidates for intervention between 18 and 26 weeks of gestation.12 The procedure conducted between 16 and 18 gestational weeks showed spontaneous loss rates of 35% up to 50%.14, 15 A 2023 study showed that neurological impairment was more significant in cases where the intervention was carried out after 23 weeks of gestation, suggesting that early intervention could reduce imbalance and positively contribute to proper neurological development,16 preventing both the death of the pump fetus and decreasing the chances of possible future sequelae.17 In the case reported, the procedure was conducted at 19 weeks and six days following the Brazilian protocol.

The main objective in the management of monochorionic diamniotic pregnancy complicated by a TRAP sequence is to guarantee the best possible outcome for the pump fetus, preserving its survival and getting as close as possible to delivery at term (\geq 37 weeks).^{4,7,12} Expectant management has a mortality rate of 50% to 75% for the pump fetus, while surgi-

cal management - indicated if there is cardiac compromise of the viable fetus, acardiac fetal mass growth, and/or polyhydramnios - provides an estimated survival rate of up to 80% for normal twins. ^{9, 1}

The appropriate method for treating the TRAP sequence is one of the main arguable points in this area. The main surgical options include interrupting blood flow through the umbilical cord to the acardiac fetus through options such as ligation with thread, photocoagulation, ablation, bipolar coagulation, sectioning the cord, or applying absolute alcohol.² Of these, the most popular seem to be intrauterine laser therapy by fetoscopy and radiofrequency ablation, which are considered safe and reliable methods.2, 12 However, none of these modalities has proven to be superior in terms of pump twin survival or minimizing complications, including negative outcomes such as premature rupture of membranes, premature birth, or unexpected death.¹⁸ In general, the choice of the technique used should consider the operator's experience, the center's resources, the clinical presentation, gestational age, and the team's preferences.1, 12 In the case described, the technique was chosen according to the experience of the fetal medicine specialist and the resources available.

Regarding the survival rate of the pump twin after the procedures, Chaveeva et al.¹⁰ found a rate of around 80% for techniques including ablation of umbilical cord vessels by laser or diathermy, coagulation of placental anastomoses by laser or ablation of intrafetal vessels by monopolar diathermy, laser, or radiofrequency. However, lower rates were found after umbilical cord ligation and the use of cord coils or intrathecal alcohol injection.¹⁰

Considering survival rates of around 80% after the laser procedure, it is recommended to terminate the gestation of the pump fetus via cesarean section in cases of poor presentation, fetal distress of the normal fetus, or other

contraindications to vaginal delivery.¹² In the case described, it was possible to conduct a successful cesarean section at 37 weeks, given the close monitoring and guarantee of maintaining fetal well-being from a cardiovascular point of view.

Therefore, given the uniqueness of the complications involving the TRAP sequence and the absence of a consensus on the most appropriate method for treatment, further studies on the subject are necessary and promising.

CONCLUSION

This paper discussed a case of successful diagnosis and the choice of the laser photocoagulation treatment technique for the abdominal insertion of the umbilical cord of the acardiac twin with the TRAP sequence. The intervention was conducted at 19 weeks and six days of gestational age, in line with the Brazilian protocol. The patient had a good post-operative evolution, and weekly ultrasounds showed a reduction in reverse blood flow compared to previous exams and the survival of the pump twin after the procedure. The route of delivery chosen by the medical team was a cesarean section at 37 weeks, which had no complications. In terms of survival of the pump fetus and reduction of possible complications, early intervention is suggested, and the main intervention methods (radiofrequency ablation and intrauterine laser therapy) are not superior when compared to each other. Generally, the center's resources, clinical presentation, gestational age, operator experience, and team preferences should be considered when choosing the method.

CONSENT

The patient has consented to having her personal information and images included in the article.

CONFLICTS OF INTEREST

The authors declare no conflicts of interest.

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