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Case Report

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Intrafetal laser therapy in acardiac twin pregnancy: a case report

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Abstract

Objective: In this study, we aimed to present the case of acardiac twin pregnancy (twin reversed arterial perfusion sequence, TRAP) successfully treated with intrafetal laser therapy.

Case: Monochorionic diamniotic twin pregnancy, a fetus without heart (acardiac fetus) and a second fetus with normal appearance (pump fetus) were observed in the anterior placenta during USG examination of the pregnant women who admitted to our clinic at her 14 weeks of gestation. Retrograde blood flow was monitored via Doppler USG and she was established with the diagnosis of TRAP. At her 15 weeks of gestation, abdominal aortic and iliac vein lines were coagulated electively by intrafetal laser procedure, and it was found that acardiac fetus was avascular, so the procedure was ended. In gestational follow-up visits, it was seen that acardiac fetus did not grow and regressed. Pump fetus development and Doppler findings had a normal progress. On the 37 weeks and 3 days of gestation, 2800 g live newborn with 8–9 Apgar score was delivered. The development of ten-month-old newborn was normal and no complication was observed.

Conclusion: Our case supports the fact that elective intrafetal laser procedure improves gestational outcomes between 12 and 16 weeks of gestation in acardiac twin pregnancies.

Keywords: Acardiac twin, intrafetal laser, management.

Özet: Akardiyak ikiz gebelikte intrafetal lazer tedavisi: Olgu sunumu

Amaç: Bu makalede intrafetal lazer tedavisi ile başarı ile tedavi edilmiş akardiyak ikiz gebelik (ikizde ters arteryel kanlanma sekansı, TRAP) olgusunun sunumu amaçlanmıştır.

Olgu: On dördüncü haftada kliniğimize başvuran gebenin ultrasonografi (USG) muayenesinde plasenta anteriyorda, monokoryonik diamniyotik ikiz gebelik, kalbi olmayan bir fetüs (akardiyak fetüs) ve normal görünümde ikinci fetüs (pompa fetüs) tespit edildi. Doppler USG ile retrograd kan akımı izlendi ve TRAP tanısı konuldu. Gebeliğin 15. haftasında elektif olarak intrafetal lazer uygulaması ile abdominal aorta ve ilyak damar hattı koagüle edildi ve akardiyak fetüsün avasküler olduğu izlendi ve işleme son verildi. Gebeliğin takiplerinde akardiyak fetüsün büyümediği ve regrese olduğu izlendi. Pompa fetüs gelişimi ve Doppler bulguları normal seyretti. Gebeliğin 37 hafta 3. gününde Apgar 8–9 olan 2800 g bebek canlı olarak doğurtuldu. On aylık olan bebeğin gelişimi normal olup, bir komplikasyon izlenmedi.

Sonuç: Bizim olgumuz da akardiyak ikiz gebeliklerde 12–16 gebelik haftaları arasında elektif intrafetal lazer uygulamasının gebelik sonuçlarını geliştirdiğini desteklemektedir.

Anahtar sözcükler: Akardiyak ikiz, intrafetal lazer, yönetim.

Introduction

Acardiac twin pregnancy (twin reversed arterial perfusion sequence, TRAP) is defined as the presence of a fetus without a heart or with a non-functional heart found in 2.6% of monochorionic twin pregnancy together with a pump fetus feeding other one through arterial anastomoses in the placenta.[1] While the mortality is 100% for acardiac fetus, the pump fetus is under the risk of cardiac failure, preterm labor and associated complications, and the mortality has been reported as 55%.[2]

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Various fetoscopic and intrafetal techniques were used to prevent the pump fetus loss and possible complications in TRAP cases. With this case report, we aimed to present acardiac twin pregnancy case which underwent intrafetal laser practice.

Case Report

Twenty-two-year-old case with gravida 1, parity 0 referred to our clinic with the diagnosis of monochorionic twin pregnancy at her 14 weeks of gestation. In her ultrasonographic examination, monochorionic diamniotic twin pregnancy, a fetus without heart (40 mm, acardiac fetus) and a second fetus (pump fetus) with 80 mm crown-rump length were observed in the anterior placenta (**Fig. 1**). Retrograde blood flow was monitored via Doppler USG and she was established with the diagnosis of TRAP. Ductus venosus flow of the pump fetus was normal. The family was informed about acardiac twin pregnancy and prognosis. Elective intrafetal laser therapy was recommended for the acardiac fetus. The family accepted the therapy one week later at 15 weeks of gestation and informed consent of the family was obtained.

Through ultrasonography, intrafetal laser therapy was carried out by using 18 G 15 cm needle. The pelvis was entered under the fetal umbilical cord of the acardiac fetus by needle, and intra-abdominal aorta and iliac vein line were reached by passing through 400 nm fiber needle. The veins on this line were coagulated by using neodymium yttrium aluminum garnet (Nd:YAG) laser source (Dornier MedTech, Munich, Germany) (**Fig. 2**). Then, the procedure was ended when it was found that acardiac fetus was avascular. Heartbeat of the pump fetus was monitored. In the follow-up a week later, no vascularity was seen in the acardiac fetus and it was found that there was no change in the dimensions. Heartbeat of the pump fetus was monitored and fetal Doppler findings were found normal. Since karyotype analysis could not be performed on the amniocentesis (AC) material, the AC procedure was carried out on the week 18 and karyotype analysis was normal. Acardiac fetus regressed in gestational follow-ups. Pump fetus development and Doppler findings had a normal progress. On 37 weeks and 3 days of gestation, cesarean section was performed with the diagnoses of oligohydramnios and fetal distress. The 2800 g infant with 8–9 Apgar score was delivered alive. The development is normal and no complication has been observed in the baby which is currently 10-month-old.

Discussion

TRAP sequence is seen in 1% of the monochorionic pregnancies. Although the pathogenesis of TRAP sequence has still been unknown, reverse blood flow from pump fetus to acardiac fetus via artery-artery anastomosis is considered as an unchanging characteristics.[1,3] Ruiz-Cordero et al.[4] investigated 13 TRAP cases in their study and reported that the interruption in early placental and embryonic vascular development is the main mechanism of abnormal hemodynamic early tissue hypoxia and the atrophy of the organs accordingly. They highlighted that single umbilical

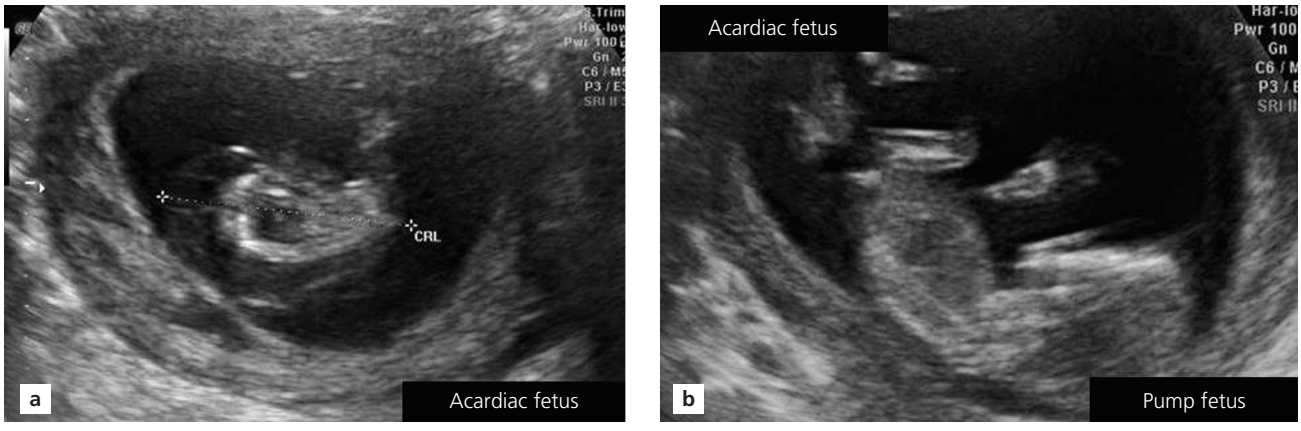

Fig. 1. Acardiac fetus (**a**), acardiac and pump fetuses (**b**).

artery, umbilical arterial thrombosis and calcification and abnormal cord insertion (velamentous cord insertion) have a key role in the development of TRAP.

Although most of TRAP cases are diagnosed in the late first and second trimesters, the prognosis is poor. The reverse arterial flow between acardiac fetus and pump fetus causes high output cardiac failure, intrauterine fetal death and polyhydramnios, all leading to preterm labor. The perfusion of acardiac fetus was stopped by various techniques to decrease high mortality rate and the complications. Fetoscopic cord ligation,[5] cord coagulation by laser,[6] alcohol injection,[7] monopolar or bipolar cord coagulation[8,9] and fetoscopic laser coagulation of placental anastomoses[10] were used to stop the flow of the acardiac fetus. Intrafetal radiofrequency ablation (RFA)[3,11] and intrafetal laser ablation[12,13] are used widely as minimal invasive techniques. Recently, it has been reported that high intensity focused ultrasound was successfully used on TRAP cases as the non-invasive technique.[14]

Lewi et al.[15] planned the procedure between 16 and 18 weeks of gestation in TRAP cases diagnosed at the first trimester and they reported the presence of spontaneous loss in pump fetus in 33% (8/24) of the cases followed up to this week, stopped spontaneous flow in acardiac fetus in 21% (5/24) of the cases and persistent flow in acardiac fetus in 46% (11/24) of the cases. One of 11 pregnancies was terminated and invasive procedure was performed in 10 pregnant women (intrafetal laser therapy in 6 cases, RFA in 1 case and laser cord coagulation in 3 cases). They reported that 9 (90%) pregnancies resulted in live birth. In the studies related with RFA use on TRAP cases, survival rate of pump fetus was reported as 71–85%.[3,11] Similarly, in cases applied intrafetal laser therapy, the survival rate of pump fetus was reported as approximately 80%.[12,13] Berg et al.[16] reported in their study that the survival rate of pump fetus was similar in RFA and intrafetal laser practices, however; preterm premature rupture of membrane (PPROM) developed in 42.9% of the pregnancies underwent RFA (<34 weeks) but PPROM was not observed in cases underwent intrafetal laser therapy. Cabassa et al.[11] reported that PPROM developed in 57% (4/7) of the cases underwent RFA. Also, in the study of Lee et al.[3] carried out on 98 TRAP cases, they reported that PPROM developed in 17% (17/98) of the cases after RFA therapy and four (4.1%) cases were lost due to PPROM and premature preterm labor during neonatal period. However, Sugibayashi et al.[17] reported that PPROM developed (<34 weeks) in 2.9% (1/35) of the cases and premature preterm labor (<34 weeks) in 8.6% (3/35) of the cases after multi-stage

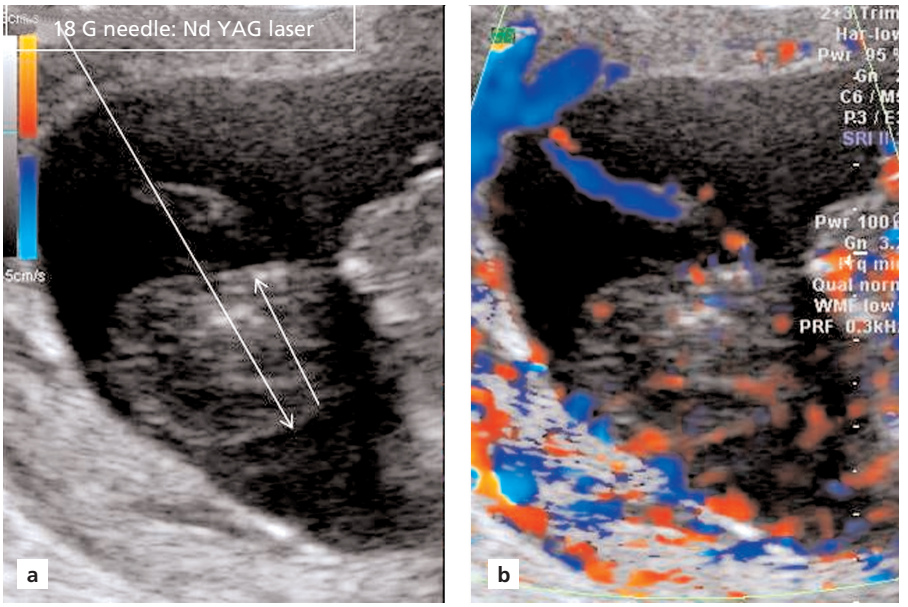

Fig. 2. Intrafetal laser practice on acardiac twin (**a**) and Doppler USG image (**b**).

RFA therapy of TRAP cases and they associated the low frequency of PPROM with the short duration of the procedure.

Although there is no consensus on the best method and time, Lewi et al. reported in their study that 33% of the siblings of acardiac twin fetuses are lost spontaneously before 16 weeks and they highlighted the necessity of performing prophylactic procedure until 16 weeks.[15] Also, Pagani et al.[12] showed in their study that poor gestational outcomes were significantly low by intrafetal laser ablation procedure before 16 weeks and they recommended performing elective intrafetal laser therapy between 13 and 15 weeks of gestation. Chaveeva et al.[13] investigated the cases who underwent intrafetal laser procedure between 12 and 27 weeks of gestation, and they could not find any correlation between therapy week and survival rate; however, they found that there was an inverse correlation between therapy week and delivery week and they reported that the elective procedure between 12 and 14 weeks of gestation might develop survival rate.

Conclusion

In conclusion, we stopped the bleeding of acardiac fetus by intrafetal laser electively at 15 weeks of gestation in our case. We found no complication in the gestational follow-ups. Our case supports the fact that elective intrafetal laser therapy between 12 and 16 weeks of gestation improves gestational outcomes.

Conflicts of Interest: No conflicts declared.

References

1. van Gemert MJ, van den Wijngaard JP, Vandenbussche FP. Twin reversed arterial perfusion sequence is more common than generally accepted. Birth Defects Res A Clin Mol Teratol 2015;103:641–3.
2. Moore TR, Gale S, Benirschke K. Perinatal outcome of forty-nine pregnancies complicated by acardiac twinning. Am J Obstet Gynecol 1990;163:907–12.
3. Lee H, Bebbington M, Crombleholme TM; North American Fetal Therapy Network. The North American Fetal Therapy Network Registry data on outcomes of radiofrequency ablation for twin-reversed arterial perfusion sequence. Fetal Diagn Ther 2013;33:224–9.
4. Ruiz-Cordero R, Birusingh RJ, Pelaez L, Azouz M, Rodriguez MM. Twin reversed arterial perfusion sequence (TRAPS): an illustrative series of 13 cases. Fetal Pediatr Pathol 2016;35:63–80.
5. Quintero RA, Reich H, Puder KS, Bardicef M, Evans MI, Cotton DB, et al. Brief report: umbilical-cord ligation of an acardiac twin by fetoscopy at 19 weeks of gestation. N Engl J Med 1994;330:469–71.
6. Ville Y, Hyett JA, Vandenbussche FP, Nicolaides KH. Endoscopic laser coagulation of umbilical cord vessels in twin reversed arterial perfusion sequence. Ultrasound Obstet Gynecol 1994;4:396–8.
7. Sepulveda W, Bower S, Hassan J, Fisk NM. Ablation of acardiac twin by alcohol injection into the intra-abdominal umbilical artery. Obstet Gynecol 1995;86:680–1.
8. Rodeck C, Deans A, Jauniaux E. Thermocoagulation for the early treatment of pregnancy with an acardiac twin. N Engl J Med 1998;339:1293–4.
9. Tan TYT, Sepulveda W. Acardiac twin: a systematic review of minimally invasive treatment modalities. Ultrasound Obstet Gynecol 2003;22:409–19.
10. Hecher K, Lewi L, Grataco's E, Huber A, Ville Y, Deprest J. Twin reversed arterial perfusion: fetoscopic laser coagulation of placental anastomoses or the umbilical cord. Ultrasound Obstet Gynecol 2006;28:688–91.
11. Cabassa P, Fichera A, Prefumo F, Taddei F, Gandolfi S, Maroldi R, et al. The use of radiofrequency in the treatment of twin reversed arterial perfusion sequence: a case series and review of the literature. Eur J Obstet Gynecol Reprod Biol 2013;166:127–32.
12. Pagani G, D'Antonio F, Khalil A, Papageorghiou A, Bhide A, Thilaganathan B. Intrafetal laser treatment for twin reversed arterial perfusion sequence: cohort study and meta-analysis. Ultrasound Obstet Gynecol 2013;42:6–14.
13. Chaveeva P, Poon LC, Sotiriadis A, Kosinski P, Nicolaides KH. Optimal method and timing of intrauterine intervention in twin reversed arterial perfusion sequence: case study and meta-analysis. Fetal Diagn Ther 2014;35:267–79.
14. Okai T, Ichizuka K, Hasegawa J, Matsuoka R, Nakamura M, Shimodaira K, et al. First successful case of non-invasive in-utero treatment of twin reversed arterial perfusion sequence by high-intensity focused ultrasound. Ultrasound Obstet Gynecol 2013;42:112–4.
15. Lewi L, Valencia C, Gonzalez E, Deprest J, Nicolaides KH. The outcome of twin reversed arterial perfusion sequence diagnosed in the first trimester. Am J Obstet Gynecol 2010;203:213.e1–4.
16. Berg C, Holst D, Mallmann MR, Gottschalk I, Gembruch U, Geipel A. Early vs late intervention in twin reversed arterial perfusion sequence. Ultrasound Obstet Gynecol 2014;43:60–4.
17. Sugibayashi R, Ozawa K, Sumie M, Wada S, Ito Y, Sago H. Forty cases of twin reversed arterial perfusion sequence treated with radio frequency ablation using the multistep coagulation method: a single-center experience. Prenat Diagn 2016;36:437–43.