CASE REPORT

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Prenatal Diagnosis of Twin Reversed Arterial Perfusion (Trap) Sequence: A Case Report

Twin Reversed Arterial Perfusion (Trap) Sekansının Prenatal Tanısı: Olgu Sunumu

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Abstract

Twin reversed arterial perfusion (TRAP) sequence is a rare condition of monochorionic twin pregnancy. It has an incidence of 1:35.000 pregnancies and constitutes 1% all monochorionic pregnancies. The etiopathogenetic mechanisms are not well defined. There is co-existence of a normal pump twin and an acardiac recipient twin. A 22-year-old nulliparous woman was referred with TRAP sequence. The prognosis in TRAP sequence is lethal for acardiac twin and the fetal mortality of the pump twin is very high due to the cardiac failure. The obstetrician should be aware of TRAP-sequence in twin/multiple pregnancies.

Keywords: Monochorionic twin pregnancy, prenatal diagnosis, sequence, twin reversed arterial perfusion (trap)

Öz

Twin reversed arterial perfusion (TRAP) sekansı monokoryonik ikiz gebeliklerde nadir görülen bir durumdur. Görülme insidansı 1:35,000 olup, tüm monokoryonik gebeliklerin yaklaşık %1'inde görülebilir. Etiyopatogenetik mekanizması tam olarak bilinmemektedir. Normal bir donör ikiz ve bir akardiyak alıcı ikiz birlikte bulunmaktadır. Olgu sunumumuzda 22 yaşında, nullipar 23. gestasyonel haftada ultrasonografik olarak tanı konulan TRAP sekansını rapor ettik. TRAP sekansı prognozu akardiyak ikiz için ölümcül olup, donör ikizde de konjestif kalp yetmezliği gelişmesi durumunda mortalite oldukça yüksektir. Kadın hastalıkları ve doğum uzmanları ikiz/çoğul gebeliklerde TRAP sekansına karşı dikkatli olmalıdır.

Anahtar kelimeler: Gebelik, monokoryonik ikiz, prenatal tanı, sequence, twin reversed arterial perfusion (trap)

Introduction

Twin reversed arterial perfusion (TRAP) sequence is a rare condition of monochorionic twin pregnancy. It has an incidence of 1:35.000 pregnancies and constitutes 1% all monochorionic pregnancies (1-3). There is co-existence of a normal pump twin and an acardiac recipient twin. The blood flows via an arterioarterial anastomosis from an umbilical artery of a pump twin into the umbilical artery of an acardiac twin and returns through a venovenous anastomosis to the pump twin. The poorly oxygenated blood from the healthy twin enters the other twin abdominally and principally perfuses the lower extremities and the body, subsequently lower concentrations of oxygen reaching the superior body parts. The upper half of the body of an acardiac twin is extremely poorly developed and sometimes not developed at all. The pump twin suffers from high output cardiac failure; therefore, it may present with cardiomegaly, ascites, pleural effusion, polyhydramnios, and skin edema (4).

The prognosis in TRAP syndromes is lethal for acardiac twin and the mortality rate for the pump twin is very high (50-75%) due to congestive heart failure (5,6).

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Case Report

A 22-year-old nulliparous woman was referred to our perinatology unit at the 23rd week of gestation with TRAP sequence. Amniotic fluid sampling was performed at the 21st week of gestation, revealing a normal 46, XX karyotype. Ultrasonographic examination showed a monochorionic monoamniotic twin pregnancy. The recipient twin demonstrated well- developed lower limbs, partially developed lumbar and thoracic spine, absent cervical spine, under-developed abdominal and chest organs (Figure 1). The heart, head and upper limbs were absent. The recipient twin was retrogradely perfused by a normal pump twin without the signs of cardiac compromise. She had a dyspnea due to the polyhydramnios, the measurement of four quadrants was calculated as 30 cm. Pelvic examination revealed a cervix 40% effaced and 0.5 cm dilated and she had uterine contractions. Tocolytic treatment was begun immediately and therapeutic amniocentesis was performed to reduce the amniotic volume and to avoid premature labor. The patient underwent corticosteroid therapy for fetal lung maturation. Fetal MR imaging and subsequent minimal invasive operative interventions were offered as alternative options, but the patient refused. However, at the 26th week of gestation, cardiomegaly, ascites and pleural effusion were demonstrated in pump twin. Therefore, a cesarean section was performed. The acardiac twin weighed 940 g and its gender was not clear (Figure 2). The pump twin weighed 850 g, was of female gender and was admitted to the neonatal intensive care unit with Apgar scores 4 and 7 at 1st and 5th min, respectively.

The histopathologic examination of the acardiac twin revealed total absence of cranial structures, cervical spines, heart, lungs, liver, gall bladder, stomach, and upper limbs. The fetus had bilateral kidneys and surrenal glands, edema of the skin, the intestine segments in omphalocele sac and lower extremities with four fingers each. The umbilical cord had two vessels. The acardiac twin was classified as holocardius (if the heart is totally absent) and acephalus (if the head is totally absent) with respect to the abnormalities (4,6). The pump twin is still alive at neonatal intensive care unit.

Discussion

The prenatal diagnosis of a complicated monochorionic multiple pregnancy and in such a case as we presented, a so-called TRAP-sequence during ultrasonographic examination is feasible and can be easily established during the first or second- trimester-screening (7,8). The etiopathogenetic mechanisms are not well defined. Although one twin is completely normal in terms of fetal anatomy, cardiomegaly and hydrops may develop secondary to cardiac overload in advanced stages. The acardiac twin most often has an underdeveloped head and upper part of body, and impressive edema involving mostly the upper body (9). An acardiac twin may be detected ultrasonographically by noticing fetal movement without

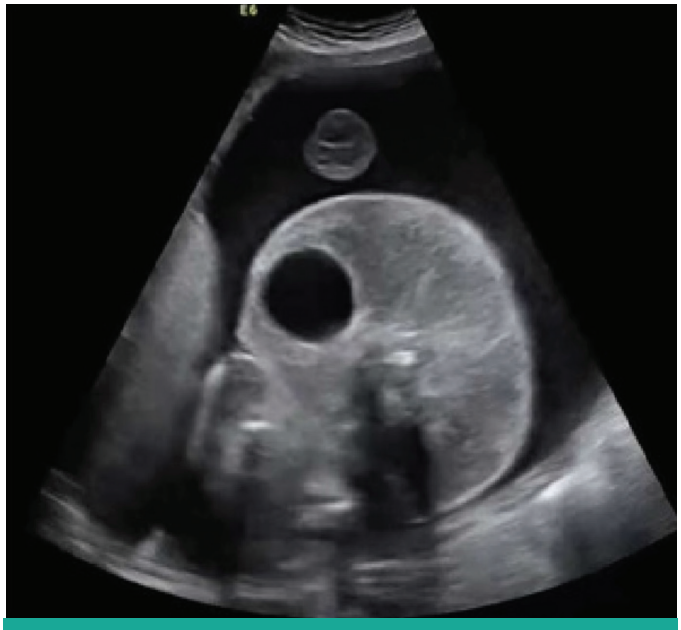

Figure 1. Ultrasonographically abdominal plane of acardiac fetus

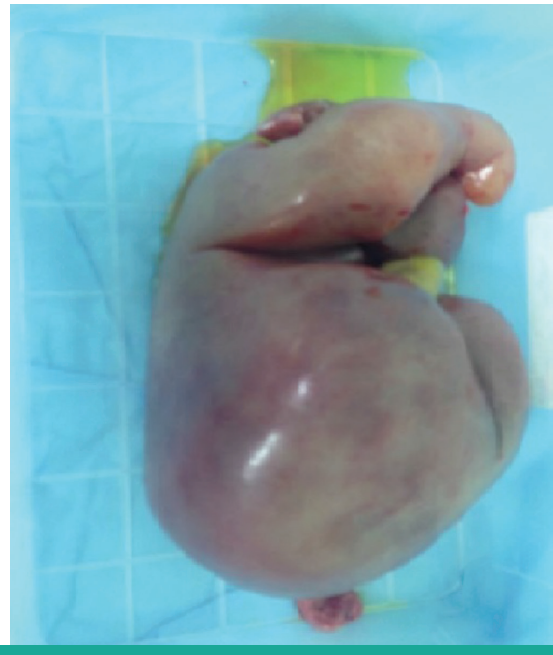

Figure 2. Macroscopic view of the acardiac fetus

a heartbeat. Doppler examination shows pathognomonic flow-pattern in terms of reversed arterial perfusion from the pumping twin towards the recipient twin. Generally, reversed arterial blood supply enters the acardiac twin via a single umbilical artery. The umbilical vessels drain usually in hypogastric or superior mesenteric artery (7,10).

In our case, there was no chromosomal and congenital anomaly. However, about one-third of the cases were reported to have an abnormal karyotype in literature (7). In addition, congenital anomalies are present in about 9% of pump twins. Potential risk of the pumping fetus is congestive cardiac failure developing due to the increased cardiac output. This high cardiac output also increases perfusion of the fetal kidneys, resulting in the overproduction of fetal urine and subsequent polyhydramnios. Preterm labor or premature rupture of membranes in TRAP-sequences may occur due to polyhydramnios. Treatment modalities include a broad spectrum of possible strategies, depending on the situation of the pumping twin. Therapeutic amniocentesis may reduce the amniotic volume to avoid premature labor. Conservative management includes different tocolytic approaches and digitalis- treatment to support the cardiac performance of the pump twin. The aim of the operative interventions is to separate the two blood-circuits of twins (7,10-15).

Ethic

Informed Consent: Written and verbal consent of the patient was obtained.

Peer-review: Externally and internally peer-reviewed.

Authorship Contributions

Follow up of the Case: Y.C., İ.Y., S.A., Literature Review: B.A., S.A., Y.C., Writing: Y.C., İ.Y., B.A., Manuscript Review and Revisation: B.A., S.A., İ.Y.

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