Unique Fetal Vascular Connections Presented in a Monochorionic Twin Placenta with Selective Intrauterine Growth Restriction

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Abstract
We present a unique case of monochorionic-diamniotic twin placenta that showed no independent circulation in the smaller twin. A 31-year-old woman visited our hospital for monochorionic twin pregnancy. At 21 weeks of gestation, selective intrauterine growth restriction and intermittent absent and reversed end-diastolic flow were observed on umbilical artery Doppler. Because of rapidly growing polyhydramnios of the smaller twin, the twins were delivered by cesarean section at 34 weeks of gestation (1780 and 1490 g). The twins showed normal growth without severe complications. The placenta had one large arterio-artery anastomosis (AAA) on its surface. Except the AAA, the arterial blood flow of the smaller twin circulated to three umbilical veins (UVs) of the larger twin. The UVs of the smaller twin were drained from the UAs of the larger twin. The smaller twin was speculated to share only 25% of the placenta.

ABBREVIATIONS
AAA: Arterio Arterianastomosis; UV: Umbilical Vein; MC: Mono Chorionic; DC: Di Chorionic; AVA: Arterio-Arterial Anastomoses; AVA: Arterio-Venous Anastomoses; VVA: Veno-Venous Anastomoses

INTRODUCTION
Monochorionic (MC) twin pregnancies are known to have higher perinatal mortality than dichorionic (DC) twin pregnancies that is attributed to the placental morphologic characteristics, especially the presence of vascular anastomoses that connect the two circulations. Placental vascular anastomoses are present in about 95% of MC twin placentas [1,2]. Three types of anastomoses have been documented: arterio-arterial anastomoses (AAA), arterio-venous anastomoses (AVA), and veno-venous anastomoses (VVA). When AVA are present, each twin individually has two parts of the MC placenta and one part that is shared and supplied by the AVA [3]. We present a unique case of monochorionic-diamniotic twin placenta with AVA and AAA that consisted of only 2 parts: a shared part and the part belonging to the larger twin. The smaller twin had no independent placenta.

CASE PRESENTATION
A 31-year-old woman, gravida 0, para 0, visited our hospital at 7 weeks of gestation because of MC twin pregnancy. At 21 weeks of gestation, an umbilical artery (UA) Doppler waveform of the smaller twin showed intermittent absent and reversed end-diastolic flow (iAREDF) (Figure 1). No abnormal pattern was detected in deep vein waveforms of both twins. The twins weighed 400 g (-0.78SD) and 323 g (-1.84SD). Amniotic fluid volumes were within normal limits for both twins. We diagnosed the smaller twin as having selective intrauterine growth restriction (sIUGR). Subsequently, we measured estimated fetal weight, UA Doppler waveforms, and DV Doppler waveforms with ultrasound once a week to determine fetal status. At 34 weeks of gestation, the larger twin weighed 1814 g (-1.26SD), with a maximum vertical pocket of amniotic fluid (MVP) of 3 cm. Meanwhile, the smaller twin weighed 1602 g (-2.11SD), with an MVP of 8.5 cm. We performed cesarean section because of rapidly forming polyhydramnios of the smaller twin. The larger twin was male, weighed 1780 g, and the Apgar score was 7 (1 min)/9 (3 min). The umbilical artery blood gas analysis of the larger twin showed pH 7.343, pCO2 41.2 mmHg, and pO2 26.5 mmHg. The smaller twin was also male, weighed 1490 g, and the Apgar score was 7 (1 min)/9 (3 min).
DISCUSSION

We experienced a unique case of monochorionic-diamniotic twin placenta that showed no independent circulation in the smaller twin. The most important determinant of discordant growth is unequal placental sharing in monochorionic twins [2]. To evaluate the placental territory of each twin, we performed color injection of the placental vasculature as previously reported in other studies [4-6]. Usually, placental share of each twin is easily determined based on the distribution of respective chorionic vessels. However, in our case, since no individual placental circulation was observed in the smaller twin on the dye injection study, it was difficult to determine the placental territories by that way. So we calculated the placental share only by counting the numbers of respective veins and ignored arteries.

We also found a large AAA in this case, which is often detected in unequally shared placentas [4]. Existence of a large AAA and large net flow reduces the impact of placental territory discordance and results in reduced birthweight discordance [3]. Therefore, these intertwin connections often play a beneficial and lifesaving role by increasing the availability of oxygen.

7 (1 min)/8 (3 min). Umbilical artery blood gas analysis of the smaller twin showed pH 7.287, pCO2 54.0 mmHg, and pO2 16.5 mmHg. The placenta weighed 746 g and measured 23 × 20 × 2.8 cm. The lengths and diameters of the umbilical cords of the larger twin were 42 cm and 2.3 cm and those of the smaller twin were 40 cm and 0.8 cm, respectively. The twins had two amnions, with the intertwin membrane that cut the center of the placenta. No macroscopic anomaly was detected in either twin. Microscopic examination revealed appropriate-appearing villi for a 35-week placenta, and apparent infarction, hemorrhage, and inflammation were not detected. Both twins were admitted to the neonatal intensive care unit and showed normal growth without severe complications. Cranial magnetic resonance imaging examination on day 24 revealed no apparent parenchymal brain lesions. Both twins were discharged from the hospital on day 31.

To evaluate the angioarchitecture and blood flow of the placenta, we performed an injection study. We cannulated a 4-Fr catheter into the veins and arteries of both cords and flushed the blood with saline. A string was placed around the vessel and was tightened around the cannula to prevent backflow and leakage of dye out of the vessel, and the vessels were then injected with colored dyes (The Davidson Marking System, Bradley Products, Inc., MS, USA). A blue dye was injected in the UA, and a yellow dye in the UV of the larger twin. Green and red dyes were injected in the UA and UV of the smaller twin, respectively. The number and type of anastomoses were noted after injection. In this study, we counted all the veins running across the placental surface, and calculated the percentage of placental territory by percentage of respective vein numbers.

The placenta after the dye injection is shown in (Figure 2). One AAA (diameter, 3 mm) was detected near the cord insertion site of the smaller twin. Except the AAA, all the arterial blood flow of the smaller twin circulated to three umbilical veins (UVs) of the larger twin (Figure 3). All UVs (35 vessels) of the smaller twin were drained from the UAs of the larger twin. No VVA was detected. There were 150 yellow vessels (UVs of the larger twin), and 35 red vessels (UVs of the smaller twin) running across the surface of the placenta. From these observations, the placental territory of the twins was calculated to be 25% for the smaller twin, and 75% for the larger twin.

![Figure 1](image1.png) Umbilical artery Doppler waveform in the smaller twin at 21 weeks. Intermittent absent (arrows A) and reversed (arrows B) end-diastolic flow (AREDIF) is noted.

![Figure 2](image2.png) The placenta after dye injection. One artery-artery anastomosis detected across the surface is 3 mm in diameter.

![Figure 3](image3.png) A schematic view of hemodynamics of the twins. No vein-vein anastomoses (VVA) detected. All umbilical arteries (UAs) of the smaller twin circulate to the umbilical veins (UVs) of the larger twin. All UVs of the smaller twin draining from the larger twin.

and nutrients to the twin who shares the smaller placenta [3]. However, different roles of large AAA have been highlighted in discordant twin studies. Gratacos et al., classified a selective sIUGR MC twin on the basis of UA Doppler flow patterns into three types that correlates with clinical behavior[6]: type I, UA Doppler with positive diastolic flow; type II, persistent absent or reversed end-diastolic flow; and type III, iAREDF. Type I has the most favorable outcome with a low risk of deterioration or unexpected fatality, and the survival rate of patients with type I flow is nearly 100%. On the other hand, patients with type III have the worst outcome, with 90% patients eventually showing deterioration or unexpected fatality and a survival rate of 60%. Patients with type II have an intermediate prognosis with a survival rate of 85% [3]. Most type II flow patients (98%) have AAA >2 mm diameter. The risks of parenchymal brain lesions in the larger twin and unexpected intrauterine fetal death of the smaller twin increase in cases with large AAA owing to the hemodynamic consequences [6]. The smaller twin in our case shared only 25% of the placenta and showed type II pattern flow on UA Doppler flow. However, the difference in body weight of the twins was 83%, and the twins grew uneventfully with no apparent neurological complications. The reason for their normal growth and the role of AAA in this case are currently uncertain.

In this case, we found polyhydramnios of the smaller twin at 34 weeks of gestation. Since no apparent fetal anomaly and placental tumor were detected, conceivable causes of polyhydramnios could be: 1) acute change in blood flow in the AAA; 2) an acute decrease in the volume of the amniotic fluid in the larger twin may have activated the renin-angiotensin system, and the circulated angiotensin may have increased urine volume of the smaller twin; and 3) an idiopathic cause. The renin-angiotensin system (RAS) is up-regulated in the donor twin and down-regulated in recipients in twin-twin translation syndrome. Furthermore, paradoxical RAS activation could occur owing to the transfer of effectors such as angiotensin II through placenta shunts [7]. Further, renin mRNA and protein expression appeared qualitatively higher in the placental territory of the recipient with twin-to-twin transfusion syndrome than in the donor and non-TTTS controls [8], and the mechanism underlying this is unclear.

We experienced a case of unique fetal vascular connections in the sIUGR MC twin placenta that showed an iAREDF pattern on UA Doppler. A precise analysis of the blood flow pattern may be useful to clarify the relationship between the outcome of the fetuses and the blood flow pattern in MC twin placenta.

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REFERENCES