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Case Report

Reverse diastolic flow of the fetal middle cerebral artery associated with placental chorangiomatosis and asymptomatic concealed placental abruption ☆

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ABSTRACT

Reverse diastolic flow of the fetal middle cerebral artery is a rare, yet ominous finding which has been associated with adverse perinatal outcomes including: intracranial hemorrhage, growth restriction, fetal-maternal hemorrhage, severe anemia, hydrops, hepatic anomaly, subsequent stillbirth, and early neonatal death. We report a case in which following notation of a nonreassuring fetal heart rate at 32 weeks' gestation, sonographic documentation of persistent reverse diastolic flow of the fetal middle cerebral artery was noted in association with sonographic findings of vascular placental dysmorphology and an asymptomatic concealed placental abruption. Subsequent fetal heart rate tracing consistent with uteroplacental insufficiency led to immediate Cesarean birth of an anemic yet nonacidotic, nonhypoxic neonate, who did well following management of respiratory distress syndrome and partial exchange transfusion. Placental abruption was confirmed at delivery. Histopathology of the placenta confirmed the presence of localized chorangiomatosis ("wandering" chorangioma). The association of reverse diastolic flow of the fetal middle cerebral artery, placental chorangiomatosis and placental abruption has not been reported previously. We conclude that in the presence of prenatal sonographic findings of placental dysmorphology and or placental abruption, insonation of the fetal middle cerebral artery should be performed to assess the possibility of increased peak systolic velocity and possible reverse diastolic flow, both associated with fetal anemia and increased likelihood of an adverse perinatal outcome.

☆ Competing Interests: The authors acknowledge that we have no conflicts of interest to report.

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Introduction

Transient or intermittent reverse diastolic flow of the fetal middle cerebral artery may reflect excessive external transducer pressure [1]. Notwithstanding, rare cases of persistent reverse diastolic flow in the fetal middle cerebral artery reflect an ominous finding, which has been associated with increased adverse perinatal outcome including: intracranial hemorrhage, growth restriction, fetal maternal hemorrhage, severe anemia, hydrops, intrahepatic bile duct malformation, subsequent stillbirth and early neonatal death [1–8]. In 2015, Brownfoot et al. reported that of 6 reported cases in the literature, fetal or neonatal death occurred in 4 of the 6 cases, and one of the survivors suffered a grade III intraventricular hemorrhage and periventricular leukomalacia [2,3]. The underlying pathophysiology of reverse diastolic flow of the fetal middle cerebral artery remains undetermined [3]. This occurrence may reflect cerebral vasoconstriction causing increased peak systolic blood flow velocities with a rapid fall off of diastolic velocities, resulting in reverse diastolic flow [3]. Alternatively, reverse diastolic flow (noted in association with anemia and low blood viscosity) may represent the combined effect of lost downstream cerebral vascular tone, blood hypoviscosity, and decreased perfusion pressure during diastole.

We present an unusual case in which following notation of a nonreassuring fetal heart rate at 32 weeks' gestation, sonographic documentation of persistent reverse diastolic flow of the fetal middle cerebral artery noted in association with sonographic findings of vascular placental dysmorphology later confirmed as chorangiomatosis and asymptomatic concealed placental abruption, led to immediate Cesarean delivery of a markedly anemic neonate, who did well following management of respiratory distress syndrome and partial exchange transfusion.

Case report

A 36 year-old P0 (blood type 0, Rh+) was followed during her first pregnancy. Following mid-trimester serum screen [low pregnancy associated plasma protein (PAPP)-A of 0.28 MoM)], which indicated an increased risk of aneuploidy (Trisomy 21), noninvasive (cell free fetal DNA) screening was negative for aneuploidy. Otherwise her pregnancy was uneventful. Due to advanced maternal age and low mid-trimester serum PAPP-A, interval sonographic estimates of fetal weight were performed and revealed a slight fall-off of growth centiles from the 51st centile at 29 weeks' gestation to the 37th centile at 32 weeks' gestation. Nonstress testing at 32 weeks' gestation was nonreactive. Fetal biophysical profile was otherwise reassuring (8/10). The patient denied having abdominal pain, uterine hemorrhage or notation of decreased fetal movements. Her blood pressure = 110/60 mmHg, heart rate = 81 bpm, and her abdomen was soft and nontender. Sonography depicted a singleton, appropriate-for-gestational age, vertex-presenting fetus with a single loop nuchal cord, with active fetal movement and an overall biophysical profile (BPP) of 8/10. No evidence of fetal hydrops was noted. The placenta appeared

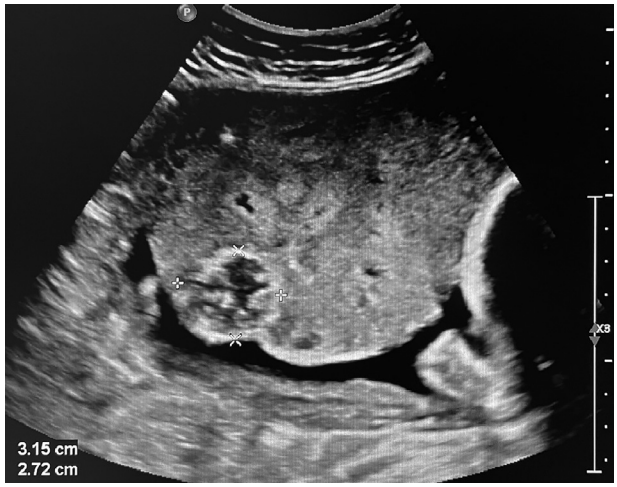

Fig. 1 – Sonographic image at 32 weeks' gestation. Note unusual structure within the placental texture. Calipers designate the measurements of the placental structure later proven at histopathology as localized chorangiomatsois (so-called "wandering" chorangioma).

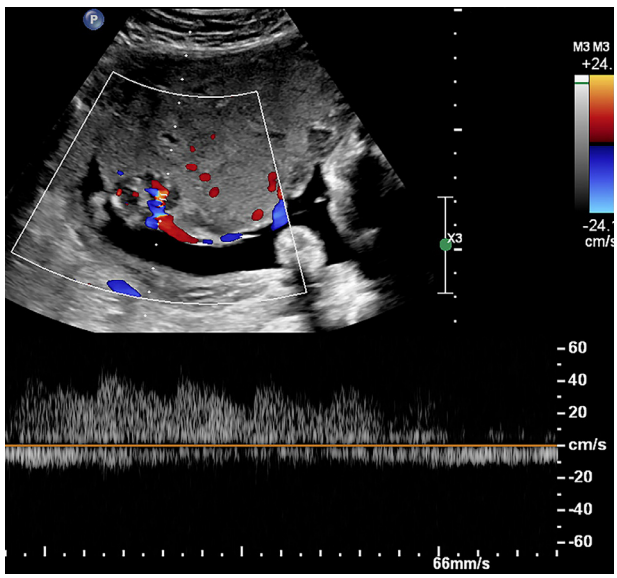

Fig. 2 – Color Doppler imaging of the later proven localized chorangiomatosis. Note the Doppler flow depicted arterial pulsations concurrent with the fetal heart rate, and venous flow.

bulky and contained vascular masses, the largest measuring 31 × 27 × 27 mm in size (Fig. 1). Color Doppler assessment of the placental vascular structures depicted arterial and venous flow (Fig. 2). In addition, an amorphous echogenic structure at the periphery of the placenta, considered consistent with a possible asymptomatic, concealed placental abruption was noted (Fig. 3). Following the notation of unusual vascular placental masses, Doppler interrogation of the fetal middle cerebral artery (MCA) was performed and depicted a peak systolic velocity (PSV) measurement of 105-131 c/sec (markedly

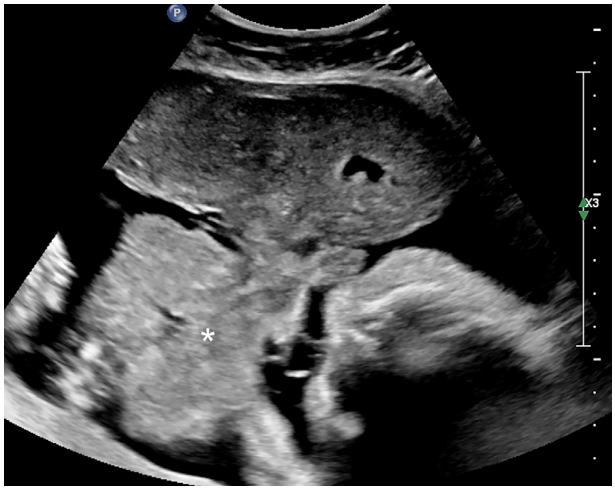

Fig. 3 – Amorphous echogenic mass noted at the edge of the placenta (marked with *) suspected to represent (and later confirmed) placental abruption.

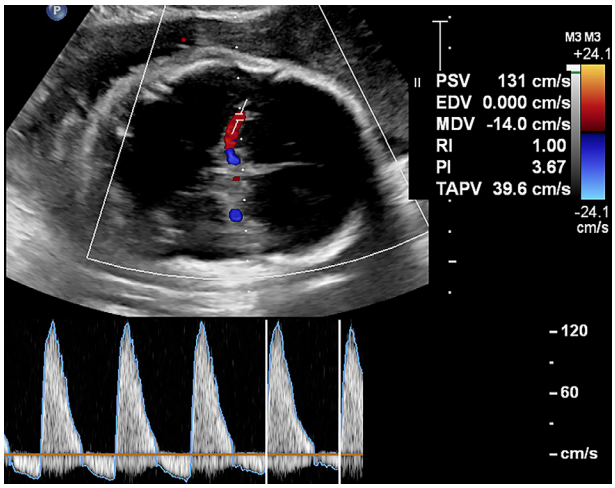

Fig. 4 – Doppler velocimetry of the fetal middle cerebral artery (MCA). Note depicted high peak systolic velocity (PSV) of 131 cm/s and reverse end diastolic flow (REDF) of this artery.

increased for gestational age), consistent with fetal anemia, and reverse diastolic flow (Fig. 4). Care was taken to ascertain that excessive transducer pressure was not applied and the above-described Doppler velocimetry findings were confirmed independently by 2 separate senior experienced sonographers. In anticipation of likely indicated preterm delivery, intramuscular antenatal corticosteroids to decrease prematurity-associated neonatal morbidities, were administered. Subsequent continuous fetal heart rate monitoring disclosed recurrent late decelerations and decreased variability, consistent with uteroplacental insufficiency, which indicated immediate delivery. At Cesarean a vigorous female neonate with Apgar scores of 6 and 8 and 1 and 5 minutes, respectively, was delivered through a transverse low uterine segment incision. A 30% placental abruption was noted. Birth weight was 1580 grams and umbilical artery Ph = 7.19 and base excess = −6.5. In the neonatal intensive care unit (NICU), the infant was noted to be considerably pale with symptomatic tachycardia and respiratory distress. At 1 hour of life, arterial pH = 7.03 and base excess = 14, indicated mixed metabolic and respiratory acidosis. With increasing respiratory distress and associated chest X-ray findings, the infant was intubated and received Surfactant. Due to symptomatic anemia with a hematocrit of 24% the infant underwent a partial exchange transfusion. Pulse oximetry indicated O2 saturation levels = 97%-99%. Post transfusion hemoglobin = 13 gr/dL and hematocrit = 39%. The infant was extubated and continued to improve clinically. During the infant's hospital course cranial ultrasound was performed and disclosed normal intracranial anatomy with no evidence of germinal matrix or intraventricular hemorrhage. The infant was discharged from the NICU on Day of life 4.

The mother's postoperative course was unremarkable. Kleihauer Betke test = 0.2, and she was discharged on postpartum Day 3. Pathology assessment of the placenta revealed a small third-trimester circumvallate placenta weighing 270 grams, with blood clots consistent with abruption. Histopathology depicted localized chorangiomatosis (so-called "wandering" chorangioma) and unremarkable 3-vessel umbilical cord. At 2 years of age, the infant remains in good health and is meeting all developmental milestones.

Discussion

Villous capillary lesions are rare abnormal placental developmental conditions which include: chorangiosis, chorangiomatosis, and chorangioma [9–12]. The causes of villous capillary lesions remain unclear, yet appear to involve excessive angiogenesis [9]. A study of 53 cases of multifocal chorangiomatosis (among 5429 consecutively accessioned placentas > 20 weeks' gestation) noted that approximately half of the placentas with multifocal chorangiomatosis exhibited avascular villi, villous chorangiosis, and distal villous immaturity. Other common placental findings included concentric narrowing of fetal villous arterioles, villous edema, and dysmorphic villi [10]. Only one case had associated placental chorioangioma [10]. Multifocal chorangiomatosis was noted associated with advanced maternal age, non-African American ancestry, nonprimigravidity, and > 5 previous pregnancies [10]. Bagby and Redline considered their findings to suggest that multifocal chorangiomatosis may represent an abnormal proliferation of the paravascular capillary net in proximal villi related to fetoplacental developmental anomalies and abnormal fetal blood flow [10]. Multifocal chorangiomatosis has been noted in association with adverse perinatal outcomes relating to severe fetal anemia [9,12]. Carlucci et al. in 2022 confirmed the association of multifocal chorioangiomatosis with unfavorable neonatal outcome. These authors described a case of severe hemolytic anemia, fetal disseminated intravascular coagulopathy, hydrops and massive umbilical vein thrombosis attributed to the presence of multifocal placental chorangiomatosis [9]. Earlier, in 2017, Arya et al. described a case of an infant born by Cesarean at

32 weeks' gestation. The neonate manifested cardiomegaly, microangiopathic hemolytic anemia, and thrombocytopenia, which resolved over time, and was attributed to diffuse placental chorangiomatosis [12]. Similarly, fetal anemia has been reported in association with placental chorioangioma [13,14].

Prenatal sonographic findings of an asymptomatic placental abruption similar to those noted in our case have been reported previously [15]. The association, if any, between multifocal chorangiomatosis and placental abruption, is unclear.

A systematic English literature search (PubMed, MEDLINE) between 1966 and 2023, utilizing the search terms "prenatal ultrasound," "placental dysmorphology," "middle cerebral artery," "reverse diastolic flow," "fetal anemia" "chorangiomatosis," and "placental abruption," confirms that the association of reverse diastolic flow of the fetal middle cerebral artery and either chorangiomatosis or placental abruption has not been reported previously. We conclude that in the presence of sonographic findings of vascular placental dysmorphology or placental abruption, Doppler interrogation of the fetal middle cerebral artery should be performed to assess the possibility of elevated peak systolic flow and possible reverse diastolic flow, both associated with fetal anemia and the increased likelihood of an adverse perinatal outcome.

Patient consent

Written informed consent for the publication of this case report was obtained from the patient.

REFERENCES

[1] Respondek M, Woch A, Kaczmarel P, Borowski D. Reversal of diastolic flow in the middle cerebral artery of the fetus during the second half of pregnancy. Ultrasound Obstet Gynecol 1997;9(5):324–9. doi:10.1046/j.1469-0705.1997.09050324.

[2] Brownfoot FC, Cluver CA, Walker SP. Persistent reversed end diastolic flow in the middle cerebral artery: an ominous finding. Ultrasound 2015;23(3):186–9. doi:10.1177/1742271X15587600.

[3] Baschat AA, Harman CR, Alger LS, Weiner CP. Fetal coronary and cerebral blood flow in acute fetomaternal hemorrhage. Ultrasound Obstet Gynecol 1998;12(2):128–31. doi:10.1046/j.1469-0705.1998.12020128.x.

[4] Sepulveda W, Shennan AH, Peek MJ. Reverse diastolic flow in the middle cerebral artery: an agonal pattern. Am J Obstet Gynecol 1996;174(5):1654–7. doi:10.1016/s0002-9378(96)70622-1.

[5] Hirshberg A, Levine LD, Schwartz N, Durnwald CP. Reversed end diastolic flow in the middle cerebral artery preceding death in a normally grown fetus. Obstet Gynecol 2013;122(2 Pt 2):507–9. doi:10.1097/AOG.Ob013e3182937854.

[6] Hjorto S, Skibsted L, Lykke JA. Reverse flow in the middle cerebral artery in a severely intrauterine growth restricted fetus. Eur J Obstet Gynecol Reprod Biol 2013;170(2):579–80. doi:10.1016/j.ejogrb.2013.07.046.

[7] De Spirlet M, Goffinet F, Phillipe HJ, Bailly M, Couderc S, Nisand I. Prenatal diagnosis of a subdural hematoma associated with reverse flow in the middle cerebral artery: case report and literature review. Ultrasound Obstet Gynecol 2000;16(1):72–6. doi:10.1046/j.1469-0705.2000.00167.x.

[8] Kawakita T, Sasaki H, Hirabuki S, Asamoto A. Fetal growth restriction and reversed middle cerebral artery end-diastolic flow with subchorionic placental lake. J Obstet Gynaecol Res 2013;39(2):578–82. doi:10.1111/j.1447-0756.2012.01994.x.

[9] Carlucci S, Stabile G, Catagini S, Borghi C, Scutiero G, Morano D, et al. Fetal disseminated intravascular coagulopathy, hydrops and massive umbilical vein thrombosis consequence of a rare placental condition: multifocal chorangiomatosis. J Matern Fetal Neonatal Med 2022;35(20):4009–13. doi:10.1080/476058.2020.1843154.

[10] Bagby C, Redline RW. Multifocal chorangiomatosis. Pediatr Dev Pathol 2011;14(1):38–44. doi:10.2350/10-05-0832-OA.1.

[11] Boroujeni AM, Yousefi E, Vincent MT, Anderson V. Chorangiomatosis: evaluation of a placental vascular lesion and relate clinical effects. Fetal Pediatr Pathol 2014;33(5-6):331–8. doi:10.3109/15513815.2014.977620.

[12] Arya S, Bhargava V, Richardson J, Hawkins HK. Diffuse chorangiomatosis as a cause of cardiomegaly, microangiopathic hemolytic anemia and thrombocytopenia in a newborn. Fetal Pediatr Pathol 2017;36(6):457–64. doi:10.1080/15513815.2017.137904.

[13] Merry-Sperry AD, Chaffin DG, Denning KL, Shamma B, Massey C, Werthammer J. Placental chorangioma with non immune hydrops. South Med J 2018;111(8):457–9. doi:10.14423/SMJ.0000000000000840.

[14] Iwahata H, Iwahata Y, Homma C, Kurasaki A, Hasegawa J, Suzuki N. Degenerative type of placental chorioangioma requiring fetal blood transfusion. J Obstet Gynaecol Res 2021;47(3):1191–4. doi:10.1111/jo.14685.

[15] Sherer DM, Kheyman M, Benayoun J, Dalloul M. Incidental sonographic finding of a concealed placental abruption leading to delivery at 37 weeks' gestation. J Clin Ultrasound 2021;49(6):630–1. doi:10.1002/jcu.22971.