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CASE REPORT

Placental Chorioangioma: A Planned Successful Outcome

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Keywords Placental chorioangioma · Case report · Polyhydramnios · Intrauterine transfusion

Introduction

Placental chorioangioma, an expansile vascular malformation, is a non-trophoblastic benign tumor originating from primitive chorionic mesenchyme. It has been noted to be the most common benign tumor of the placenta [1], being diagnosed as an incidental finding. Conversely, large-sized tumors (> 4 cm) have adverse fetal and maternal outcomes.

Case Report

A 26-year-old, primigravida, 31 weeks + 5 days of gestation, referred to SRIHER for an early delivery. Conception being spontaneous, pregnancy was confirmed with urine beta HCG at 45 days of amenorrhea. Scan done at 12 weeks showed an intrauterine gestation corresponding to dates, placenta forming anteriorly. First trimester screening was negative.

Target scan done at 21 weeks ruled out anomalies. Placenta was growing anteriorly with a 6.5 × 5.2-cm-sized placental mass near the cord insertion site. It showed hypoechoic echogenicity and color flow mapping showed vascularity—diagnosed as chorioangioma. Hydramnios was noted: single deepest pocket (SDP) = 9.1 cm. Fetus showed mild cardiomegaly, prominent aorta with normal myocardial function. Fetal Doppler and echocardiography was normal. There were no signs of fetal anemia or cardiac failure. Distance between umbilical cord insertion site and chorioangioma was 3.7 cm. Feeding vessel diameter was 5.5 mm.

Weekly ultrasonography with Doppler study and fetal echocardiography was done to monitor growth of the placental mass, liquor status and signs of circulatory changes in the fetus. Gradual increase in size of the mass and liquor level was noted.

Scan done at 26 weeks showed increase in mass from 115 cc to 160 cc. Liquor: SDP = 11.5 cm. Fetal cardiomegaly with minimal pericardial effusion was noted. Asymmetry of cardiac chambers (RT > LT) with prominent aorta was found. Doppler study showed middle cerebral artery peak systolic flow (MCA PSV) 1.7 MoM suggestive of fetal anemia along with pulsatile turbulent flow in portal vein, engorged hepatic veins with multiple cystic channels communicating with the hepatic veins suggestive of hyperdynamic circulation.

In view of the above, intrauterine transfusion was planned for the patient. 35 ml of packed cells (O negative blood group) was transfused to the fetus through the umbilical vein at the placental insertion site. At 26 weeks, two doses of Inj. Betamethasone (12 mg) were given 24 h apart.

One week post transfusion, at 27 weeks + 6 days, scan showed placental chorioangioma (167.2 cc), SDP = 10.9 cm. Fetal weight estimated at 1055gm with mild cardiomegaly and tricuspid regurgitation. Doppler study showed MCA PSV 1.42 MoM, ductus venosus—high resistance flow and hyperdynamic circulation. In view of an uptrend of MCA PSV, patient was planned for ultrasound-guided ablation of the feeder vessel of the chorioangioma. 4 ml of 20% lipiodol solution mixed with 1 ml of histoacryl glue solution was flushed with 5% dextrose solution. 2.5 ml of the above solution was injected into the feeder vessel. Cessation of

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blood flow was noted within 5 min along with blanching of chorioangioma. Post-embolization, MCA PSV: 1.21 MoM and there was no evidence of significant fetal anemia.

On day 26 post procedure at 31 weeks and 5 days, the chorioangioma had increased in size (450 cc) and vascularity (120 cc) with polyhydramnios (SDP 12.9 cm). Fetus showed cardiomegaly, mild tricuspid along with mitral regurgitation and bilaterally compressed lungs. Doppler showed MCA PSV 1.49 MoM with prominent portal system (umbilical and portal vein showing pulsatile flow) and hyperdynamic circulation. In view of the above changes, she was referred to SRIHER for an early delivery.

On admission, her vitals were stable. Per abdomen uterus was overdistended, relaxed. Nonstress test was reactive.

She delivered at 31 weeks + 5 days, by lower segment cesarean section (breech extraction)—male baby, weighing 1.56 kg. Apgar score at 1 min and 5 min was 6/10 and 8/10, respectively. Intraoperative period was uneventful. Placenta and membranes were delivered in toto. Baby had a weak cry at birth and was intubated after 2 cycles of positive pressure ventilation. Following 2 weeks of stay in neonatal intensive care unit, baby and mother were discharged at 21 days. Baby weighed 1.83 kg, on paladai feeds at discharge.

On gross examination, placenta showed a 20 cm × 12 cm × 5 cm homogenous, mahogany-colored, well-circumscribed, globular mass on the fetal surface, situated 4 cm from site of cord insertion. Maternal surface showed congested vessels (Fig. 1). No areas of hemorrhage/infarction were noted.

Discussion

Despite a low incidence, placental chorioangiomas are the most common benign placental tumors, noted more in multiple pregnancies and female babies. It is believed to form by 16th day of fertilization; however, there has been no known documentation in the first 12 weeks.

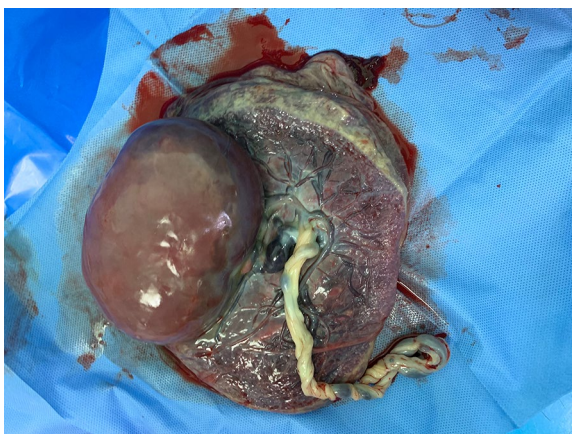

Fig. 1 Placental chorioangioma specimen

While small-sized tumors are clinically insignificant and missed on routine ultrasonography, large-sized tumors (> 4 cm) are easily diagnosed. Ultrasound with Doppler remains the gold standard for diagnosis and differentiating it from other placental masses (placental teratoma, degenerated myoma, partial hydatidiform mole and placental hemorrhage). It is usually found on the fetal surface of the placenta, near umbilical cord insertion. Magnetic resonance imaging (MRI) is used only in doubtful cases, whereas computed tomography (CT) has minimal role to play due to high radiation risk and poor tissue differentiation. CT scan has no role in detecting metastasis as chorioangiomas are always benign.

Maternal complications are preeclampsia, preterm labor, placental hemorrhage, polyhydramnios, malpresentation and postpartum hemorrhage. Of the reported complications, the correlation of chorioangioma with polyhydramnios is significant. Among fetal complications, non-immunologic hydrops, hemolytic anemia, congenital anomalies, fetal thrombocytopenia, cardiomegaly and growth restriction have been noted. Fetal congestive heart failure, as in this patient, may develop due to increased blood flow through the low resistance vascular channels in the chorioangioma acting as an arteriovenous shunt. Doppler ultrasound with serial fetal echocardiography on a weekly basis can be used for diagnosis of fetal cardiac failure, like it was done in this case. Most cases reported are associated with adverse perinatal outcome. The size of the mass and the presence of fetal hydrops are likely to be the main determinants of perinatal outcome [2].

Three histological patterns of chorioangioma have been classified by Marchetti: angiomatous, cellular and degenerative. Immunohistochemically, the tumor cells show staining for CD31, CD34, factor VIII, GLUT1 and cytokeratin [3].

Treatment modalities for large chorioangiomas aim at temporary measures for prolonging gestation. These include polyhydramnios—serial therapeutic amnioreduction along with maternal indomethacin and fetal anemia—intrauterine transfusion. Betamethasone before 34 weeks of gestation is recommended for fetal lung maturity.

Based on the fetal/maternal complications, several in utero interventions have been proposed—fetal transfusions, endoscopic surgical devascularization, alcoholic ablation, interstitial laser treatment, microcoil embolization, bipolar electrosurgery and, if gestational age permits, early delivery.

In this case, early detection of chorioangioma, close follow-up and monitoring was done using serial ultrasounds (Fig. 2) and Doppler studies. Follow-up is targeted at monitoring the vascularity and growth of the chorioangioma as well as progressive fetal compromise. Due to regular monitoring, gestation could be prolonged and fatal complications alleviated with timely use of intrauterine fetal

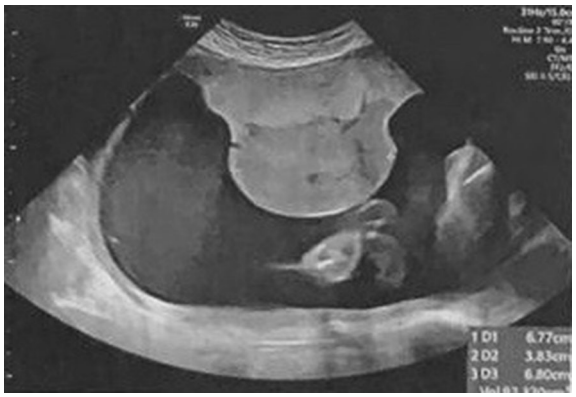

Fig. 2 Ultrasound image of chorioangioma

transfusion and embolization of the feeder vessel. This highlights the possibility of a healthy fetal and maternal outcome with a systematic follow-up.

While the above invasive modalities have been described in the literature, previously published cases have limited themselves to tocolysis, amnioreduction and early delivery in view of imminent preterm labor. Hence, there is limited data and experience on the same.

Conclusion

Placental chorioangioma is a rare case in the field of obstetrics; there are lack of management guidelines/treatment protocols. The above is a case where timely intervention and prompt institutional delivery led to a successful feto-maternal outcome. Ultrasonography with Doppler remains the gold standard for diagnosis and follow-up. Invasive techniques—embolizaton and fetal transfusion—used to prolong gestation, despite minimal experience with the same, have proved to have a successful outcome.

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Compliance with Ethical Standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical Approval This article does not contain any studies with human participants or animals performed by any of the authors.

Informed Consent Informed consent was obtained from the patient mentioned in the case report. Consent was obtained from the patient for publication of this case report.

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Tanvi Desai is a third year resident in the department of OBGYN at Sri Ramachandra Institue of Higher Education and Research, Chennai. She completed her MBBS at D.Y. Patil School of Medicine, Navi Mumbai with a gold medal in Pediatrics. Post MBBS she gained clinical experience in the field of Gynecologic Pathology at Mount Sinai, New York (2017) and was awarded the Glickman fellowship in Gynecologic Oncology at Weil Cornell, New York (2017). She received the Young Turks' Award by FOGSI and won 3rd place in the poster presentation at World Conference on Conception to Delivery, Mumbai (2019). Her current thesis topic is in Urogynecology and she aspires to specialize in laparoscopic gynecology.