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CASE REPORT

Placental Chorioangioma: A Rare Cause for Polyhydramnios and Fetomaternal Complications

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ABSTRACT

Aim: Placental chorioangioma is a rare cause for polyhydramnios which is often missed among its differential diagnosis, leading to grave fetomaternal complications. This case report aims to provide insight, that, despite their rarity, placental tumors are a potential cause for fetomaternal complications.

Background: Chorioangioma is a nontrophoblastic benign vascular tumor leading to maternal complications and fetal complications. Timely diagnosis and intervention help improve fetomaternal outcomes.

Case description: A 29-year-old, second gravida at 29 weeks 5 days gestation was admitted to labor room with preterm labor pains, history of a gush of watery discharge per vaginum, severe respiratory embarrassment, and bilateral lower limb edema. On per abdomen examination, liquor was increased, and the abdomen was distended up to xiphisternum. Ultrasonography revealed hyperechoic lesion approximately 6.6 x 4 cm arising from the placenta, suggestive of chorioangioma placenta. She delivered vaginally a live male baby weighing 1.9 kg. The baby had respiratory distress and was kept in neonatal intensive care unit on continuous positive airway pressure (CPAP) mode of ventilation. The woman had a bout of postpartum hemorrhage necessitating blood transfusion. Placental examination revealed lobular purplish red growth attached with a pedicle to the fetal surface of the placenta. Histological examination showed an angiomatous pattern of chorioangioma. As she presented late, we had no scope of providing conservative management for better fetomaternal prognosis.

Conclusion: Adverse fetomaternal outcome in case of large chorioangioma warrants timely diagnosis and intervention.

Clinical significance: Though rare, placental tumors must be considered as a differential diagnosis in cases of polyhydramnios.

Keywords: Nontrophoblastic placental tumors, Placental chorioangioma, Polyhydramnios, Preterm labor.

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BACKGROUND

Placental chorioangioma, a nontrophoblastic benign vascular tumor of the placenta, is among one of the rare causes for polyhydramnios with the incidence of 0.6–1%, which, may lead to adverse fetomaternal outcomes. It is the most common type of placental tumor.[1] Small tumors (<5 cm) are usually asymptomatic, but large tumors may be associated with grave feto maternal complications.

We present a case, with chorioangioma of placenta resulting in polyhydramnios, preterm, and adverse fetomaternal outcome.

CASE DESCRIPTION

A 29-year-old, unbooked patient, second gravida at 29 weeks 5 days of gestation presented to us with a history of a gush of clear fluid per vaginum since one day. She also complained of dyspnea, vague pain abdomen, and abdominal distension for 2 weeks. She had respiratory embarrassment and bilateral lower limb edema (not subsiding on rest) since one week. She had no history of increased blood pressure (BP). Her blood sugar recordings were normal. Her blood group was O negative.

On examination, the patient was afebrile, pulse rate 86 per minute, BP 120/70 mm Hg in right arm in sitting position, jugular venous pressure (JVP) normal, respiratory rate 36 per minute. She had severe dyspnea which aggravated on lying down. Her cardiovascular and respiratory system examination was normal. She had the bilateral pedal and pretibial pitting edema. There was no evidence of jaundice, anemia, cyanosis or clubbing. On per abdomen examination, fundal height was more than the period of gestation with abdomen overdistended up to xiphisternum. Liquor was increased. The uterus was irritable. Abdominal girth was 40 inches. Fetal heart rate was 150 beats per minute, regular. On speculum examination, clear fluid was seen leaking

from OS. On per vaginum examination, os was 1.5 cm dilated membranes ruptured, fetal head felt at the station-2 and active leaking present. Her routine investigations were normal. Glucose tolerance test was also normal. Indirect Coomb's test was negative.

Ultrasonography (USG) showed a single live fetus with cephalic presentation corresponding to 30 weeks of gestation with polyhydramnios (AFI:30 cm) and single deepest pocket of 18 cm. The placenta was on the posterior wall upper segment, grade II. A heterogeneous hyperechoic lesion measuring approximately 6.6 × 4 cm was seen arising from the placenta, bulging into amniotic cavity and closely abutting the insertion of cord to placenta suggestive of chorioangioma. Extensive internal vascularity was noted on doppler. Estimated fetal weight was 1.703 kg + –255 g (Fig. 1). Doppler velocimetry of umbilical artery showed normal low resistance spectral waveform with the good forward diastolic flow with S/D ratio of 2.7. Fetal middle cerebral arterial (MCA) showed normal high resistance spectral waveform with S/D ratio of 4.1. There was no evidence of fetal hydrops or any gross structural abnormalities.

She had spontaneous preterm labor and delivered a live male baby, weighing 1.9 kg with Apgar scores 4 and 6 at 1 and 5 minutes, vaginally, same day. She had a bout of atonic PPH with estimated blood loss of around one liter which was managed conservatively with uterotonics and blood transfusions. Her estimated blood loss was around one liter. The baby had respiratory distress and was urgently shifted to neonatal, intensive care unit (NICU) and was put on CPAP mode of ventilation. There was evidence of neonatal sepsis which was managed empirically with ampicillin and gentamicin which was later stepped up to injection (inj) meropenem on account of constantly rising c-reactive protein (CRP) levels. Later blood C/S revealed the growth of *Candida* species and inj fluconazole was then added. After two weeks, the baby recovered. The baby did not have any evidence of hydrops or any gross structural abnormalities. Her estimated blood loss was around one liter.

On gross examination of the placenta, the placental disc measured 16 × 4 × 5 cm and weighed 2 kg. The fetal surface of the placenta showed prominent blood vessels and a lobular growth, purplish red, which measured approximately 6 cm × 5.5 cm × 3.5 cm and attached to the fetal surface of the placenta with a pedicle. Cut surface of the mass was purplish red (Fig. 2). Histological examination of placenta showed an angiomatous pattern of chorioangioma (Fig. 3). The adjacent placenta showed features of chorioangiosis with microcalcifications.

DISCUSSION

Nearly 1% of all pregnancies are complicated by polyhydramnios. Among the various common causes leading to polyhydramnios are central nervous system and gastrointestinal tract anomalies, fetal isoimmunization and maternal diabetes mellitus. As one tends to focus on the above fetal and the maternal causes, a rare cause of polyhydramnios, i.e., placental tumors may be missed. The primary neoplasms of the placenta may be classified into two groups: trophoblastic and nontrophoblastic. Nontrophoblastic diseases are more common, always benign and comprise of chorioangioma and teratoma which can originate from any part of the placenta except trophoblastic tissues.[2-4] The incidence of chorioangioma is in 0.6–1% of placentas. Histologically, chorioangiomas may be classified into angiomatous (capillary), cellular, and degenerative types. The capillary type is the most common histological subtype.[5] Advanced maternal age, diabetes, hypertension, female sex of the fetus, premature labor, first delivery, and multiple pregnancies are common associated underlying risk factors. Small chorioangiomas

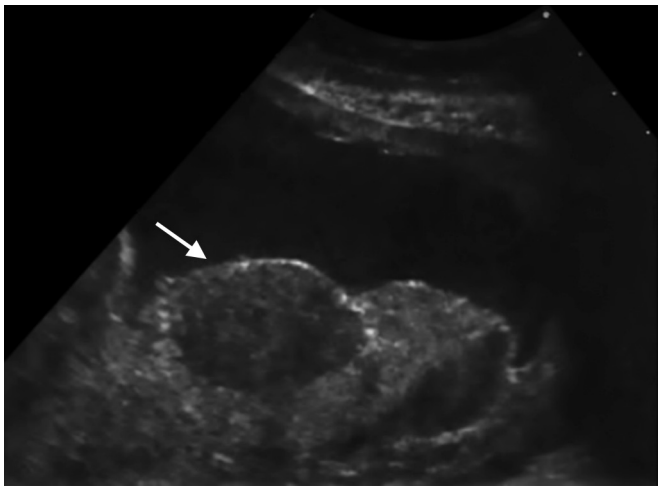


Fig. 1: Ultrasonography showing placenta on the posterior wall with a heterogeneous hyperechoic lesion measuring approximately 6.6 × 4 cm seen arising from the placenta suggestive of chorioangioma

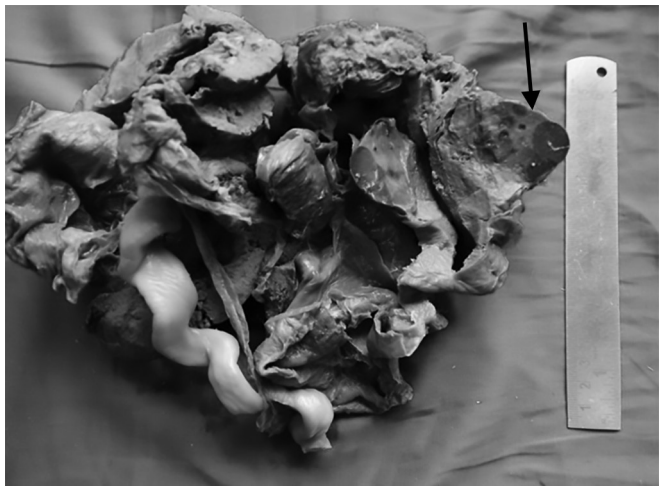
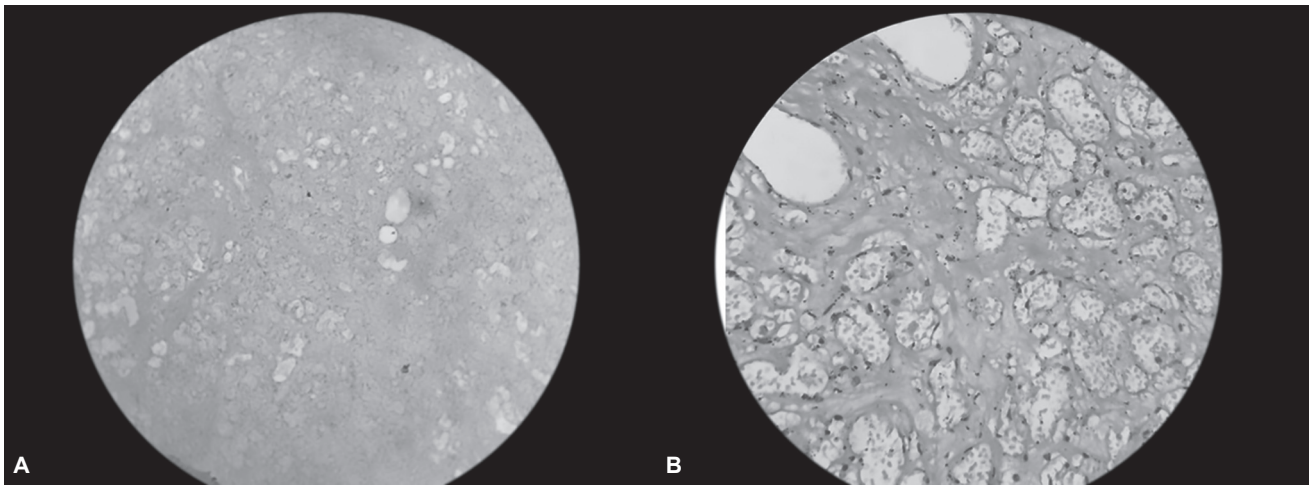


Fig. 2 : Gross examination of the placenta showing the placental disk measuring 16 × 4 × 5 cm, weighing 2 kg and site of placental chorioangioma (arrow) measuring approximately 6 cm × 5.5 cm × 3.5 cm



Figs 3A and B: Histological examination of placenta showing an angiomatous pattern of chorioangioma: (A) HPE x 10X; (B) HPE x 40X

are usually missed on routine ultrasonography and are not associated with fetal or maternal complications. Larger masses (those >5 cm), are easily diagnosed by ultrasound scan and cause feto-maternal complications.[6]

Maternal complications of placental chorioangioma include polyhydramnios, premature rupture of membranes, cervical incompetence, premature labor; placental abruption; malpresentation; increased risk of cesarean section; and postpartum hemorrhage.The formation of polyhydramnios in these cases may be explained by several theories. First, increased intravascular pressure caused by obstruction of the blood flow by a tumor near the umbilical cord insertion may increase transudation into the amniotic cavity. Second, the large surface area of the enlarged vessels of the angioma may also predispose to the increased transudation. Third, partial placental insufficiency caused by shunting of the fetal blood into the vessels of the chorioangioma may also lead to polyhydramnios.

Fetal complications include fetal congestive heart failure, thrombocytopenia, nonimmunologic fetal hydrops, hemolytic anemia, intrauterine growth restriction, brain infarction, umbilical vein thrombosis, fetal cerebral embolism, and intrauterine fetal and neonatal death.[7]

Chorioangioma is believed to arise as early as the 16th day of fertilization. It is usually found on the fetal surface of the placenta usually near umbilical cord insertion.[8] The gold standard in primary diagnosis is Doppler USG which aids fetal monitoring. At USG, chorioangioma appears as a well-defined complex echogenic mass different from the rest of placenta and protruding into amniotic cavity near umbilical cord insertion. It often mimics placental teratoma, degenerated myoma, and blood clot. Doppler USG helps differentiate between the three. On Doppler, in chorioangioma, feeding vessel usually has the same pulsatile flow as that of the umbilical artery but may have arteriovenous shunt causing low resistance flow. Echo pattern of blood clot differs with time, while chorioangioma remains the same. Partial mole has the diffuse pattern, and myoma is seen in the maternal surface. Rare differential diagnosis of chorioangioma also includes chorangiosis and chorangiomatosis. Suspicious cases can be diagnosed by magnetic resonance imaging. Immunohistochemically, the tumor cells show staining for CD31, CD34, factor VIII, GLUT1 and cytokeratin. Treatment modalities of chorioangioma include serial fetal transfusions, endoscopic surgical devascularisation, alcoholic ablation, and interstitial laser coagulation. Polyhydramnios is treated with therapeutic amniocentesis and maternal indomethacin therapy. Steroid administration before 34 weeks is recommended for fetal lung maturity.

This patient had a large, highly vascular tumor associated with serious fetomaternal complications. The patient had polyhydramnios leading to preterm labor, PPROM, and preterm vaginal delivery. Owing to uterine overdistention, she had atonic Postpartum hemorrhage requiring blood transfusions. She was an unbooked case and had not undergone any antenatal work up. She presented to us for the first time in an emergency in the third trimester with spontaneous preterm labor and PPROM owing to which we had no scope of providing her conservative management for better fetomaternal prognosis.

The patient and baby were discharged in stable condition. On follow-up visits to the postnatal clinic, she is now doing well.

CONCLUSION

Large chorioangioma being associated with adverse fetomaternal outcomes warrant institutional and timely delivery. Antenatal diagnosis is by ultrasound, and Doppler is the investigation of choice inaccurate diagnosis of chorioangioma. Careful visualization of the placenta on

ultrasonography in cases of polyhydramnios is required as chorioangioma is one of the rare cause of polyhydramnios. The timely diagnosis helps improve fetomaternal outcomes. Regular follow-up helps in timely diagnosis and intervention.

CLINICAL SIGNIFICANCE

Though placental tumors are rare, they should be considered as differential diagnosis in cases of polyhydramnios.

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