International Journal of Reproduction, Contraception, Obstetrics and Gynecology
Agrawal P et al. Int J Reprod Contracept Obstet Gynecol. 2023 Apr;12(4):1168-1171
www.ijrcog.org
pISSN 2320-1770 | eISSN 2320-1789

DOI: https://dx.doi.org/10.18203/2320-1770.ijrcog20230836

Case Report

Giant placental chorioangioma presenting as severe polyhydramnios: a case report

Prabha Agrawal[1]*, Rahul Agrawal[2], Shabnam S. Fareed[1], Kinnera R. Ancha[3]

[1]Department of Obstetrics and Gynaecology, [2]Department of Internal Medicine, [3]Department of Anaesthesiology, Care Hospital, Hyderabad, Telangana, India

Received: 12 February 2023
Accepted: 09 March 2023

***Correspondence:**
Dr. Prabha Agrawal,
E-mail: drprabha1001@yahoo.com

ABSTRACT

Chorioangiomas are the most common non-trophoblastic, benign, vascular tumour of the hemochorial placenta. Small chorioangiomas are usually symptomless, and of no clinical significance while giant ones more than 4 cm in diameter may be complicated by polyhydramnios, foetal cardiomegaly, hydrops fetalis, and foetal growth restriction. We present a case of a 32-year-old primigravida referred to us at 30 weeks of gestation with large placental chorioangioma causing polyhydramnios which was treated by amnioreduction twice over 1 month. On referral the tumour size was about 56 mm size with severe polyhydramnios with amniotic fluid index of 57 cm, with breathlessness and pain abdomen. After relevant investigations and informed consent, she was taken up for caesarean section. 2 litres of clear liquor drained. She delivered a live female baby weighing 1.2 kg with Apgar score of 7 and 8. Patient stood the operation well. Gross and microscopic examination of the placenta confirmed the diagnosis of chorioangioma. Chorioangioma should be considered as differential diagnosis in cases of hydrops fetalis or polyhydramnios. Doppler ultrasound is the method of choice to detect chorioangioma and its vascularity. Giant chorioangiomas complicating pregnancy can be managed conservatively with close surveillance, foetal monitoring and timely intervention to prevent maternal and foetal morbidity and mortality.

Keywords: Chorioangioma, Polyhydramnios, Pregnancy

INTRODUCTION

Primary placental tumours are divided into trophoblastic and non-trophoblastic tumours. Trophoblastic tumours consist of molar pregnancy, placental site trophoblastic tumour and choriocarcinoma. Non-trophoblastic tumours consist of chorioangioma and teratoma.[1] Chorioangiomas were first described by Clarke in 1798.[2] They are benign vascular tumours of placenta, majority being single, small and tend to occur on the foetal side of the placenta. They are the most common type of placental tumour with an estimated incidence of 1% of all pregnancies.[2] Chorioangiomas are malformations formed as a result of defective angiogenesis and exhibit higher proliferation rate than tumour free placental tissue.[3]

Large chorioangiomas more than 4-5 cm, are rarely seen in obstetric practice with an approximate incidence of 1 in 10,000 pregnancies.[4] Increased incidences is seen in twin pregnancies, maternal hypertension, diabetes, and female foetal sex.[4] In majority of cases, small chorioangiomas are asymptomatic, and treated with expectant management. Large size chorioangiomas are more often diagnosed prenatally and, are clinically significant due to poor outcomes for both the foetus and the mother like polyhydramnios, foetal heart failure, hydrops fetalis, intrauterine growth restriction (IUGR) and intrauterine foetal death. In view of the associated high perinatal death rate (30–40%), a number of therapeutic interventions have been introduced with various outcomes such as direct interventions to the chorioangioma and drainage of the excess amniotic fluid.[5]

CASE REPORT

A 32-year-old primigravida was referred to our hospital at 30 weeks of gestation with severe shortness of breath (SOB), pain abdomen, severe hydramnios and a placental tumour, for further management. She had history of pain abdomen and SOB for 4 weeks. She had no significant past medical, surgical or family history. The oral glucose tolerance test done at the 24th week of gestation was negative. In view of pain abdomen, the ultrasound done at 26 weeks revealed a hypoechoic area in the central portion of the placenta measuring 42×20×20 mm with increased vascularity. Her amniotic fluid index (AFI) was 41 cm, with no signs of structural abnormalities or foetal hydrops. Her foetal growth and biophysical profile were normal. Under proper asceptic precautions ultrasound-guided amnioreduction procedure was performed, successfully aspirating 1 litre of clear fluid resulting in decrease of pain abdomen and tightness. Follow-up ultrasound examination after 2 weeks showed a gradual increase in AFI to 48 cm. Two intramuscular injections of betamethasone 12 mg were given 24 hours apart for foetal lung maturity. Again ultrasound-guided amnioreduction procedure aspirating 1 litre of clear fluid was performed resulting in clinical improvement.

However, at 30 weeks there was gradual increase in the intensity of uterine contractions and SOB so she was referred to our hospital in view of tertiary care with neonatal facilities. On admission, the vital signs were in normal range. Her haemoglobin was 10.4 gm/dl with random blood sugar of 96 mg/dl and serum thyroid stimulating hormone level of 3.2 mIU/l. She was perceiving the foetal movements well. The uterus was tense, contracting and over distended to a size corresponding to 36 weeks of gestation. Her ultrasound examination showed AFI of 57 cm with an ovoid intra-placental mass protruding into amniotic cavity of 56×51×48 mm likely chorioangioma. Umbilical artery doppler showed normal end diastolic flow and middle cerebral artery peak systolic velocity (MCA PSV) was normal for gestational age.

Following multidisciplinary team discussion, caesarean section was done under spinal anaesthesia. 2 litres of clear liquor drained. She delivered a live female baby weighing 1.2 kg with Apgar score of 7 and 8. Patient stood the operation well. Approximate blood loss was 500 ml. On gross examination of the placenta, a single well-circumscribed firm, tan-brown mass of 5.6 cm diameter was seen on the placenta (Figure 1).

Baby was shifted to neonatal intensive care unit (NICU). Patient was discharged in a stable condition after 3 days and baby was shifted out of NICU after 7 days. Microscopic examination of placenta showed histological features consistent with chorioangioma with a three-vessel umbilical cord. Circumscribed tumour was seen with small capillary sized channels lined by single layer of endothelial cells and stroma composed of stromal cells and collagen. Scattered mitosis were present up to 15/10 high power field (Figure 2). Weight of the placenta was 520 grams. There was no evidence of nuclear atypia or malignancy. The tumour was confirmed to be atypical chorioangioma. On follow-up, both the patient and her child were well.

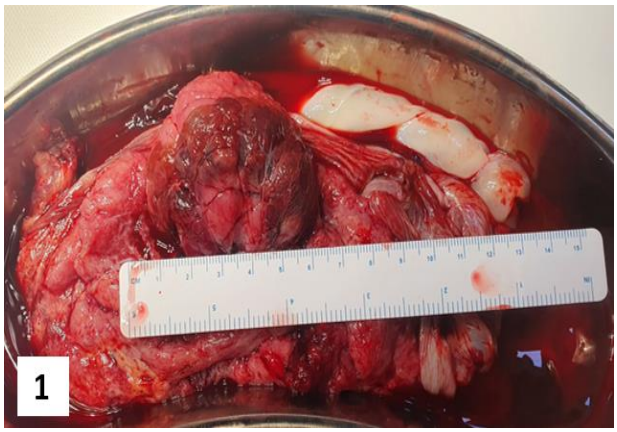

Figure 1: Gross appearance of the placenta with a single, well-circumscribed, tan-brown chorioangioma.

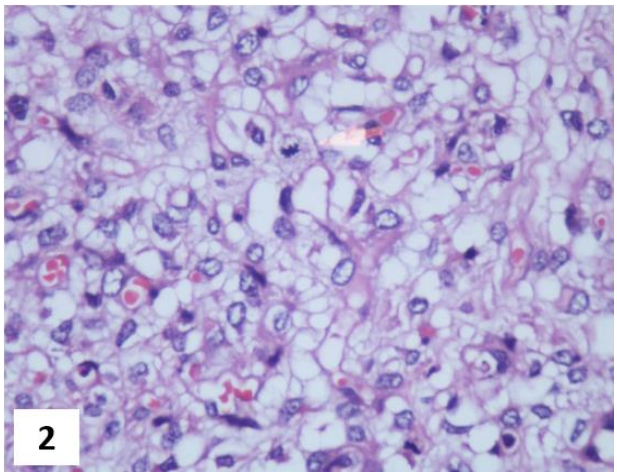

Figure 2: Microscopic picture (H&E, 40x) showing circumscribed tumour with capillary like channels and scattered mitosis.

DISCUSSION

Large or multiple chorioangiomas have been variably associated with a number of foetal complications including anaemia, thrombocytopenia, nonimmune hydrops, cardiomegaly, congestive cardiac failure, IUGR, Beckwith-Wiedemann syndrome, single umbilical artery, umbilical vein thrombosis, foetal cerebral embolism and intrauterine death.[2,6] Unexplained foetal tachycardia is a known presentation of chorioangioma.[4] Maternal complications include polyhydramnios, cervical incompetence, premature labour, placental abruption, pre-eclampsia, malpresentation; increased risk of caesarean section, postpartum hemorrhage, hepatic and cutaneous hemangiomas and maternal mirror syndrome.[2,6,7]

A retrospective study done by Zanardini et al in a tertiary foetal medicine unit over a 7-year period (2001 to 2007) showed that out of 19 cases of giant placental chorioangioma, two-thirds of the cases developed complications related to foetal growth restriction or tumour-related effects that required either delivery or intervention.[5]

Several theories have been postulated to explain the mechanism of polyhydramnios like, direct transudation into the amniotic fluid and increased foetal urine production secondary to the foetal hyperdynamic circulation and maternal mirror syndrome (generalized fluid overload and preeclampsia). AFI of 5 to 25 cm is considered normal. Polyhydramnios (AFI ≥25 cm), can be further classified into mild (25-30 cm), moderate (30.1-35 cm), or severe (>35 cm).[8] Hyperdynamic circulation caused by shunting of the foetal blood into the vessels of the chorioangioma may cause foetal high-output cardiac failure, hydrops fetalis and uteroplacental insufficiency. Foetal anaemia has been ascribed to microangiopathic hemolysis, foetomaternal hemorrhage and hemodilution (due to sequestration of red blood cells and platelets by the tumour).

Doppler ultrasound examination is the gold standard for diagnosis of chorioangioma. Magnetic resonance imaging is used infrequently. Placental chorioangioma is typically seen on ultrasound as a well-defined hypoechoic mass bulging from the placental surface and consisting of solid and cystic components.[1] On the basis of the blood flow within the mass, colour doppler imaging can be used to differentiate vascular tumour from hematoma.[1] Maternal and foetal complications and prognosis are directly related to, proximity of the chorioangioma to the cord, tumour's size and vascularity.[9] Other tests include foetal echocardiography for assessment of cardiac function and measurement of foetal MCA PSV. Increased MCA-PSV, pulsatile umbilical venous flow velocity and abnormal foetal echo suggest foetal anaemia. Serum levels of alpha-fetoprotein (AFP) can be raised in cases of placental angioma but is not pathognomonic due to physiological and pathological increase in other foetal and maternal medical conditions.[6]

Asymptomatic small chorioangiomas are usually treated with expectant management with a careful monitoring with serial ultrasound every 6-8 weeks. Large tumours require follow-up ultrasound examinations every 1-2 weeks to monitor growth of the tumour, heart function, MCA PSV and amniotic fluid volume. Therapeutic amnio-drainage can be done when the deepest amniotic fluid pocket is ≥12 cm accompanied by breathlessness and tense abdominal distension. Based on maternal symptoms approximately 1.5 to 3 litres of amniotic fluid can be reduced, but it usually recurs limiting the efficacy of procedure.[8] Foetal blood transfusion can be done if the foetus is suspected to be anaemic on MCA-PSV doppler assessment.[4] The use of amnio-drainage and intrauterine blood transfusion are well established for temporary relief of symptoms, but they do not correct the primary pathology. A number of other treatment modalities have been published including foetoscopic laser coagulation, bipolar electrosurgery, intra-tumoral alcohol injection and microcoil and enbucrilate embolization of feeding vessels.[7,10]

During a 10-year study (2001 to 2011) by Gruca-Stryjak et al, 7 cases of the placental tumour were prenatally detected. In 2 cases of giant chorioangioma [8-10 cm] intrauterine transfusion was performed to correct foetal anaemia and both babies had no hemodynamic problems at birth.[1]

The decision to deliver the patients must be dictated by the gestational age of onset, foetal maturation and the type, severity and progression of maternal and foetal complications. Criteria to deliver the patients is based on the presence of one of the following: reversed umbilical artery end-diastolic flow (UA-EDF) from 32 weeks, absent UA-EDF from 34 weeks, ductus venosus pulsatility index (DV-PI) above the 95th percentile before 32 weeks, significant deterioration of the foetal biophysical profile (≤6/10) or foetal heart rate short-term variation less than the 2.5th percentile.[5]

Differential diagnosis is from subamniotic hematoma, partial hydatidiform mole, placental teratoma, chorioangiocarcinoma and submucosal uterine fibroid.

CONCLUSION

Although the placental tumours are rare, they should be considered in cases of polyhydramnios or hydrops fetalis. Giant chorioangiomas complicating pregnancies may be managed conservatively with persistent observation and monitoring until delivery is necessary. In cases where progressive polyhydramnios or signs of foetal cardiac failure are observed, clinical intervention is mandatory to prevent foetal hypoxia or demise.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

REFERENCES

1. Ropacka-Lesiak M, Gruca-Stryjak K, Breborowicz G. Nontrophoblastic placental tumors. Neuro Endocrinol Lett. 2012;33(4):375-9.
2. Weerakkody Y, Yap J. Placental chorioangioma. Radiopaedia. Available at: https://radiopaedia.org/articles/placental-chorioangioma. Accessed on 11 November 2022.
3. Guschmann M, Henrich W, Dudenhausen JW. Chorioangiomas--new insights into a well-known problem. II. An immuno-histochemical investigation of 136 cases. J Perinat Med. 2003;31(2):170-5.

4. Willis C, Ferguson S, Soydemir F. Placental chorioangioma associated with polyhydramnios and hydrops fetalis. BMJ Case Rep. 2019;12(1):e227828.
5. Zanardini C, Papageorghiou A, Bhide A, Thilaganathan B. Giant placental chorioangioma: natural history and pregnancy outcome. Ultrasound Obstet Gynecol. 2010;35(3):332-6.
6. Abdalla N, Bachanek M, Trojanowski S, Cendrowski K, Sawicki W. Placental tumor (chorioangioma) as a cause of polyhydramnios: a case report. Int J Womens Health. 2014;6:955-9.
7. Vaidyanathan G, Bai A, Perchani J, Said HE, Lakhtakia R. Placental chorioangioma: usual presentation and alternate treatment options. J Case Rep. 2017;30-2.
8. Hwang DS, Mahdy H. Polyhydramnios. In: StatPearls. Treasure Island (FL): StatPearls Publishing. 2022.
9. Panat V. Chorioangioma: A Case Report. J. Fetal Med. 2020;7:325-9.
10. Babic I, Tulbah M, Kurdi W. Antenatal embolization of a large placental chorioangioma: a case report. J Med Case Rep. 2012;6:183.

Cite this article as: Agrawal P, Agrawal R, Fareed SS, Ancha KR. Giant placental chorioangioma presenting as severe polyhydramnios: a case report. Int J Reprod Contracept Obstet Gynecol 2023;12:1168-71.