

Giant Placental Chorioangioma with Severe Polyhydramnios and Foetal Anaemia- A Case Report

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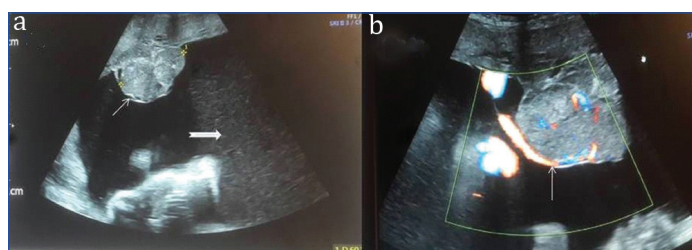
ABSTRACT

Giant placental chorioangioma is a rare, benign, vascular hamartoma of the placenta of >4-5 cm in size, since size beyond this range is associated with adverse perinatal outcome. This is a case of giant placental chorioangioma with severe polyhydramnios and foetal anemia. Serial ultrasound surveillance was able to detect foetal anemia due to the arterio-venous shunt in the chorioangioma. Severe polyhydramnios was managed with repeated amnio-reduction to relieve the mother. A non-reassuring cardiotocography and suspicion of placental abruption, following the last amnio-reduction led to an emergency caesarean. Perinatal morbidity was increased due to severe foetal anemia, coagulopathy and prematurity which was successfully managed.

Keywords: Amnioreduction, Doppler, Middle cerebral artery, Ultrasound

CASE REPORT

A 36-year-old, para 2, lady was referred at 28 weeks to the foetal medicine department with the diagnosis of severe polyhydramnios and placental chorioangioma. Grey scale ultrasound with color doppler imaging confirmed a well-circumscribed hypoechoic placental mass [Table/Fig-1] projecting into the amniotic cavity, close to the cord, measuring 61×57×47 mm, with both arterial and venous blood flow as well as severe polyhydramnios (single deepest pocket of 20 cm). The maternal abdomen was excessively distended (abdominal girth 120 cm). Apart from the great discomfort, all other maternal parameters including blood pressure and glucose tolerance test was normal. An appropriate for age grown foetus with no anomaly or hydrops. Doppler study of middle cerebral artery ruled out anaemia at the initial presentation.



[Table/Fig-1]: Ultrasound picture of giant placental chorioangioma. a) Grey scale image showing chorioangioma (thin white arrow) along with increased amniotic fluid (thick white arrow). b) Colour Doppler showing the vascularity of the chorioangioma (white arrow).

Amnio-reduction was done to relieve the mother of excessive abdominal discomfort from severe polyhydramnios. Amniotic fluid analysis did not reveal any foetal chromosomal abnormality. On a repeat scan 5 days later, MCA PSV at 95th centile (1.55 MOM) suggested foetal anemia. At this stage, laser ablation to devascularize the chorioangioma, with its attendant risks was offered to the patient, which she declined. She was also given a course of antenatal steroids due to the threat of preterm labour.

Ongoing foetal surveillance with ultrasound confirmed recurrence of severe polyhydramnios necessitating repeat amnioreduction again at 30 weeks (AFI 56.4) and 33+6 weeks (AFI 33.6). Meanwhile, the foetal MCA PSV suggested foetal anemia (MCA PSV 1.62 MOM-1.48 MOM) without signs of hydrops or cardiac failure. Following the last amnioreduction, emergency caesarean was undertaken due to suspicion of placental abruption and a non-reassuring foetal heart trace pattern.

Intraoperatively, the foetus was in transverse lie with blood stained liquor. Approximately, 3L of liquor and 700 mL of blood loss was estimated. Macroscopic examination and histology of placenta confirmed the diagnosis of giant placental chorioangioma measuring 9×7×6 cm. The placenta weighed 900 gms. The baby was born pale, with apgar of 2, 6, 8 (1, 5, 10 min) and needed neonatal resuscitation. At birth, the cord (venous) gases were 7.26 BE -4.4, lactate 5.6 mmol/l and weight 2375 grams. Fetal haemoglobin was 7.4 g/dL and the baby had bleeding from stump and nasogastric tube. The baby was transfused 17 packed red cells, fresh frozen plasma and cryoprecipitate and was referred to a higher centre where she was treated for anaemia, coagulopathy, jaundice and hypoglycaemia. Haemolytic anaemia and sepsis were ruled out. She was discharged after 21 days. At one year follow-up, baby was well with no apparent neurodevelopmental delay.

DISCUSSION

The reported incidence of giant chorioangiomas is 0.29%-0.11% [1,2]. They are associated with a perinatal mortality rate of about 40% [3-5]. Foetal complications include anemia, thrombocytopenia, non-immune hydrops, cardiac failure, growth restriction and foetal demise [3]. The central factor for the foetal complications with this vascular tumour is that it acts as an arterio-venous shunt. The fetoplacental blood volume is increased and leads to increase venous return, foetal cardiomegaly, hydrops and heart failure [1]. The hyperdynamic circulation could eventually lead to foetal demise as can be in acute events like rupture of the vessels within the chorioangioma [1,3]. This shunt can compromise the functional capacity of the rest of the placenta and cause uteroplacental insufficiency leading to its association with foetal growth restriction and foetal demise [3]. Maternal complications include polyhydramnios leading to preterm labour, abnormal lie, premature rupture of membranes, placental abruption, and emergency caesarean and post-partum haemorrhage [1,3].

The index case presented with severe polyhydramnios due to the giant placental chorioangiomas. Polyhydramnios is the most common complication of giant placental chorioangioma and is attributed to three reasons: increased foetal urine production due to the hyperdynamic circulation, increased transudation from the abnormal tumour vasculature or increased transudation from the umbilical vein in the cord due to the mechanical compression by the commonly adjacent tumour [1,3]. In this case, the tumour was also

located close to the cord. Repeated amnio-reduction was done in this patient for maternal relief due to severe polyhydramnios. However, following the third amnioreduction, placental abruption occurred which increased the foetal morbidity by adding fetomaternal haemorrhage and prematurity to the neonatal complications.

Kim A et al., also reported placental abruption with multiple rapid amnioreductions [6]. Some reports have shown that amnioreduction should be performed following vascular occlusion of feeder vessels to reduce the likelihood of further foetal deterioration [7,8]. Amnioreduction in the presence of a compromised foetal cardiac status may worsen the foetal status due to increase in the arteriovenous shunting from the reduction in the intra-amniotic pressure [1,3,4] as could have happened in this case with the last amnioreduction. This implies that the decision for amnioreduction should be judiciously taken keeping in view the risks of the procedure and the prior haemodynamic status of the foetus. Medical therapy with sulindac (cox2 inhibitors) for treating polyhydramnios has been reported at 26 weeks by Al Wattar BH et al., though it was not for severe polyhydramnios [9].

Severe foetal anemia in this case could be explained due to the dual elements of chronic sequestration of blood in the chorioangioma and placental abruption. Foetal anemia and thrombocytopenia also results from microangiopathic haemolysis within the hamartomatous capillaries [1,3,10]. However, there was no haemolysis in the baby in this case. The foetus in the present case also had coagulopathy which could be due to the same mechanism as explained in Kasabach-Merritt Syndrome where multiple microthrombi form within the tumour leading to consumptive coagulopathy in utero [10,11].

The role of ultrasound with color Doppler imaging is indispensable in a case of giant placental chorioangioma. It also helps to differentiate from other placental masses (haematoma, teratoma) as well as in prognosticating because increased vascularity of the chorioangioma is associated with more adverse outcomes [1,2] and spontaneous thrombosis with better outcomes [3]. It is an essential modality to detect the foetal complications of anemia, heart failure and growth restriction.

The choice of prenatal intervention depends upon the gestation age, the foetal cardiac status and the resources. Amnio-reduction can be done in severe polyhydramnios best after devascularisation of the chorioangioma due to risks of placental abruption and worsening status of an anemic foetus as experienced by us. It is better to do amnioreduction after. Intrauterine transfusions to treat foetal anaemia/or hydrops, may be needed once or more depending upon gestation. Definitive treatment by devascularising the tumour has been done by various techniques with the highest success rate reported with laser coagulation by interstitial technique [12]. Other modalities as bipolar electro surgery, clip application, injection of absolute alcohol and embolisation with micro coil or enbucrilate have also been reported. One needs to be wary of the risks to the foetus which is dependent upon the technique, vascular map of

the chorioangioma, position of the placenta, proximity to the cord insertion as well as status of foetal compromise [1,3]. Risks of foetal exsanguinations, collateral damage and foetal demise need to be explained to the mother.

CONCLUSION

The placenta should thus be evaluated critically in all case of polyhydramnios. Giant placental chorioangioma is a potentially high risk condition for both foetus and mother and its diagnosis entails follow-up with ultrasound in the antenatal period for foetal complications. The need, timing and type of any prenatal intervention requires individualisation with preparedness for acute foetal and maternal complications.

Abbreviations

MCA: Middle cerebral Artery

PSV: Peak Systolic Velocity

MOM: Multiple of Median

AS: Apgar Score

AFI: Amniotic Fluid Index

REFERENCES

- [1] Fan M, Skupski DW. Placental chorioangioma: Literature review. *J Perinat Med.* 2014;42:273-79.
- [2] Ulkumen BA, Pala HG, Nese N, Baytur Y. Prenatal diagnosis and fetomaternal outcomes of two cases with placental chorioangioma. *Case Reports in Obstetrics and Gynecology* Volume 2013, Article ID 926743, 3 pages <http://dx.doi.org/10.1155/2013/926743>.
- [3] Zanardini C, Papageorgiou A, Bhide A, Thilaganathan B. Giant placental chorioangioma: Natural history and pregnancy outcome. *Ultrasound Obstet Gynecol.* 2010;35:332-36.DOI: 10.1002/uog.7451.
- [4] Lim FY, Coleman A, Polzin W, Jaekle R, Habli M, Van Hook J, et al. Giant chorioangiomas: Perinatal outcomes and techniques in fetoscopic devascularization. *Fetal Diagn Ther.* 2015;37(1):18-23. doi: 10.1159/000363600. Epub 2014 Aug 28.
- [5] Merry-Sperry AD, Chaffin DG, Denning KL, Shamma B, Massey C, Werthammer J. Placental chorioangioma with nonimmune hydrops Fetalis. *South Med J.* 2018;111(8):457-59. doi: 10.14423/SMJ.0000000000000840.
- [6] Kim A, Economidis MA, Stohl HE. Placental abruption after amnioreduction for polyhydramnios caused by chorioangioma. *BMJ Case Rep.* 2018;2018. pii: bcr-2017-222399. doi: 10.1136/bcr-2017-222399.
- [7] Jauniaux E, Ogle R. Color doppler imaging in the diagnosis and management of chorioangiomas. *Ultrasound Obstet Gynecol.* 2000;15:463-7.6
- [8] Mendez-Figueroa H, Papanna R, Popek EJ, Byrd RH, Goldaber K, Moise KJ Jr, et al. Endoscopic laser coagulation following amnioreduction for the management of a large placental chorioangioma. *Prenat Diagn.* 2009;29:1277-78.
- [9] Al Wattar BH, Hillman SC, Marton T, Foster K, Kilby MD. Placenta chorioangioma: A rare case and systematic review of literature. *J Matern Fetal Neonatal Med.* 2014;27(10):1055-63.
- [10] Abiramalatha T, Sherba B, Joseph R, Thomas N. Unusual complications of placental chorioangioma: Consumption coagulopathy and hypertension in a preterm newborn. *BMJ Case Rep.* doi:10.1136/bcr-2016-215734.
- [11] Gultekin ND, Yilmaz FH, Altunhan H, Findik S, Tokgoz H, Caliskan U. Coexistence of Kasabach-Merritt Syndrome and placental chorioangioma in a premature infant. *Journal of Neonatal-Perinatal Medicine.* 2018;11(2):209-13. doi:10.3233/npm-181754.
- [12] Papaioannou GK, Evangelinakis N, Kourtis P, Konstantinidou A, Papantoniou N. Giant chorioangioma treated with interstitial laser coagulation. *Ultrasound Obstet Gynecol.* 2018;52 (2):280-81. doi: 10.1002/uog.18941. Epub 2018 Jul 4.

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