CASE REPORT

Placental chorioangioma associated with polyhydramnios and hydrops fetalis

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SUMMARY

A 27-year-old multigravida woman was noted on routine growth scan at 27 weeks gestation to have a central placental hypoechoic area measuring 6.7×6.0×4.4 cm. A subsequent magnetic resonance scan confirmed a solid mass in the placenta lying anteriorly; therefore, a preliminary diagnosis of giant placental chorioangioma was made. A repeat ultrasound scan at 30 weeks gestation indicated that the mass had increased, with the presence of polyhydramnios. The patient experienced reduced fetal movements at 31 weeks gestation. There was persistent fetal tachycardia at 33 weeks gestation, and consequently the neonate was delivered by emergency caesarean section. The placenta revealed a large chorioangioma. The neonate's birth weight was 2.85 kg and non-immune hydrops fetalis was diagnosed. The neonate improved significantly in the neonatal intensive care unit and is currently well with no medical problems.

BACKGROUND

Chorioangiomas are benign, non-trophoblastic, vascular neoplasms of the placenta.¹ They are the most common histological type of placental tumour, with an estimated incidence of 0.6%.² Incidence is increased in maternal hypertension, diabetes, twin pregnancies and female fetal sex.³ Most commonly, chorioangiomas are small and symptomless, with negligible clinical significance.⁴ Giant chorioangiomas, greater than 4–5 cm in diameter, are rare, affecting approximately 1 in 10 000 pregnancies.⁵ Giant chorioangiomas are more easily detected by ultrasound scanning and are associated with adverse perinatal and fetal outcomes. Maternal complications include intrauterine growth restriction, preterm labour and polyhydramnios. Fetal complications include haemorrhage, hydrops fetalis and mortality.⁶ The use of Doppler ultrasound has proven helpful in determining chorioangioma vascularity, with increased vascularity related to an increased risk of fetal complications.¹

CASE PRESENTATION

A 27-year-old multigravida with a history of caesarean section and normal delivery was diagnosed with gestational diabetes at 13+3 weeks gestation. The patient had a body mass index of 46.2 and had gestational diabetes in the previous pregnancies. At 27+5 weeks, a routine growth ultrasound scan demonstrated a hypoechoic area in the central portion of the placenta with increased

vascularity, measuring 6.7×6.0×4.4 cm. Umbilical artery Doppler was performed, with normal end diastolic flow. Maternal vital signs were normal, fetal movements were present and the estimated fetal weight was on the 50th centile. A magnetic resonance scan was arranged 2 days later (28+0), confirming a solid mass in the placenta lying anteriorly (figure 1). Large vessels were present within the mass in close proximity to the umbilical cord. There were no overtly malignant characteristics to the mass; therefore, a preliminary diagnosis of chorioangioma was made.

A repeat ultrasound scan performed at 30+5 weeks indicated polyhydramnios. The mass in the placenta had increased in size to 8.1×7.3×4.8 cm. Growth and fetal movements remained normal. However, at 31+5 weeks, the patient presented with reduced fetal movements. Ultrasound scan did not suggest significant changes and the cardiotocogram (CTG) trace was reassuring. At 33+1 weeks, the patient presented for a repeat CTG, which demonstrated persistent fetal tachycardia of unknown cause. Maternal vital signs were normal. Following multidisciplinary team (MDT) discussion, a category III emergency caesarean section was performed.

Figure 1 An MRI T2-weighted image illustrating a well-defined mass in the placenta (red arrows). Low-to-intermediate signal intensity was identified with some focal high signal change. Large vessels are present within the mass, in close proximity to the umbilical cord.

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A female neonate weighing 2850 g (95th centile) was delivered, with Appearance, Pulse, Grimace, Activity, Respiration (APGAR) scores of 6 and 9 at 1 and 5 min. Following delivery, the neonate was noted to be grunting, tachycardic and mildly hydropic. Haemoglobin levels from the umbilical cord blood were 98 g/L, indicating anaemia. Direct Coombs test was negative. Non-immune hydrops fetalis was diagnosed. The neonate was intubated and admitted to the neonatal unit to receive a blood transfusion and intravenous antibiotics.

INVESTIGATIONS

Macroscopic histopathological analysis revealed an irregularly shaped placenta measuring 23×18.5× up to 4 cm depth. The placenta was above the 90th centile for gestation. A single well-circumscribed mass was identified with a smooth encapsulated surface, measuring 10.5×9.5×7 cm, within which was a dominant yellow-coloured nodule of approximately 3 cm diameter. Large areas of the mass were infarcted with loss of nuclear basophilia and some areas of the convexity were covered with syncytiotrophoblasts. Overall, the appearance was consistent with a giant chorioangioma. Small foci of fibrin deposition were noted under the chorionic plate, which raised the possibility of high blood pressure as a cause. There was no evidence of maternal or fetal vasculopathy.

DIFFERENTIAL DIAGNOSIS

Differential diagnoses included chorangiocarcinoma.

OUTCOME AND FOLLOW-UP

By day 4 in the neonatal unit, the neonate was spontaneously ventilating in air, haemoglobin levels had stabilised and intravenous antibiotics were discontinued. The patient recovered well from the caesarean section with no postoperative complications. On follow-up, both the patient and her child are well. The child has no medical problems and requires no medications.

DISCUSSION

The approach to cases of chorioangioma must be dictated by both fetal maturation and maternal and fetal well-being. Treatment of chorioangioma remains controversial. Treatment options are available, such as drainage of the excess amniotic fluid and interventions to the chorioangioma itself including laser coagulation and electrosurgery.⁷ However, these interventions all pose risks and after MDT discussion were not performed in our case.

Polyhydramnios is a frequent complication of chorioangioma. One possible explanation of the polyhydramnios noted in this case was the close proximity of the tumour to the umbilical cord. There is potential that the chorioangioma compressed the umbilical vessels leading to fluid accumulation.⁴ Hydrops fetalis, defined as the abnormal accumulation of fluid in fetal interstitial compartments, is a rare complication of chorioangioma. The underlying pathology may relate to shunting of fetal blood volume through the chorioangioma.⁴ Generally, hydrops fetalis is associated with poor outcomes and fetal demise. However, we have presented a case with a favourable fetal outcome.⁴ Unexplained fetal tachycardia is a recognised presentation of chorioangioma.⁸ When combined, the manifestation of hydrops fetalis and fetal tachycardia is suggestive of fetal cardiac failure. Therefore, the presence of the advanced neonatal resuscitation team at delivery was a necessity in this case.

In this case, MRI was used antenatally to observe for malignant characteristics of the mass and exclude a more suspicious cause. Expert histopathological examination was necessary following delivery to exclude chorangiocarcinoma, an extremely rare malignant placental tumour.⁹ In chorangiocarcinoma, there is proliferation of atypical trophoblasts within the chorioangioma mass, with the potential to cause metastases within the mother and neonate.

In summary, giant placental chorioangiomas are associated with an increased risk of fetal demise. Therefore, we emphasise the need to consider the diagnosis of chorioangioma with all clinical presentations of hydrops fetalis and polyhydramnios. Additionally, we stress the importance of regular fetal heart rate monitoring in cases of suspected chorioangioma.

Patient's perspective

I was very scared and anxious when I had the ultrasound scan that revealed the chorioangioma, and even more anxious when I had the MRI scan. I was a little relieved when I was told that the Doctors believed it wasn't cancerous, but I still didn't know what to expect or anticipate. I was in a lot of pain towards the end of my pregnancy. I couldn't walk very far at all without bursting into tears and the excess fluid made me feel as though I was going to pop. I couldn't feel my daughter's movements at all, hence why I was often coming into the day unit for monitoring and I'm so glad I was otherwise the chorioangioma might not have been picked up in time! I wasn't expecting to deliver my baby at 33 weeks, it was another very scary experience. The c-section went very calmly and smoothly considering it was an emergency, I was pleasantly surprised. I later found out that I had almost 10 litres of amniotic fluid and it was actually quite comical in theatre as the surgeons told me that they had never seen so much fluid! It was horrible when my daughter was whisked away to NICU but I knew she was in the best hands. She was very swollen and puffy with the hydrops fetalis, but this soon resolved. She is now 10 months old and doing great, you would never believe she was born with these problems or born prematurely, she is coming on leaps and bounds. She has no medical conditions and does not need any medications. I am truly grateful for everyone who helped my daughter and I... you saved her life.

Learning points

- ▶ The approach to cases of chorioangioma must be dictated by both fetal maturation and maternal and fetal well-being.
- ▶ The diagnosis of chorioangioma must be considered in fetal presentations of hydrops fetalis or polyhydramnios.
- ▶ Giant chorioangiomas complicated with hydrops fetalis may be managed conservatively with regular observation and fetal monitoring until delivery is necessary.
- ▶ Regular fetal heart rate monitoring is required in all suspected cases of chorioangioma.
- ▶ Expert histopathological analysis is required to exclude chorangiocarcinoma.

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