Idiopathic Chronic Fetomaternal Haemorrhage Resulting in Hydrops – A Case Report

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Abstract

***Introduction**: We report a case of idiopathic chronic fetomaternal haemorrhage (FMH) that developed in the late trimester. **Clinical Presentation**: The patient presented with decreased fetal movement at 38 weeks gestation. Antenatal follow-up was uneventful with normal serial ultrasound performed at 22 and 35 weeks. Prior to delivery, the cardiotocography (CTG) was abnormal with decreased baseline variability and late deceleration. Emergency lower segment caesarean section was performed. Upon delivery, a hydropic neonate with a haemoglobin level of 3.9 g/dL was noted. The Kleihauer-Betke test was positive, confirming FMH. **Outcome**: The neonate later developed intraventricular haemorrhage (IVH) and spastic cerebral palsy on follow-up. **Discussion**: It is possible for FMH to occur late at the third trimester leading to detrimental effect. The fact that FMH can occur without antecedent risk factors underscores the importance of further research, and a high index of suspicion.*

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Key words: Cerebral palsy, Fetal anaemia, Kleihauer-Betke test

Introduction

Small amounts (<0.1 mL) of fetal blood are commonly found in maternal circulation.[1] Massive fetomaternal haemorrhage (FMH) involves fetal blood loss into the maternal circulation of greater than 150 mL or more than half the fetal blood volume.[2] Most cases of acute and chronic fetomaternal haemorrhage are of unknown cause, most often spontaneous and involve uncomplicated near-term pregnancies.[3,4] A literature review by Giacoia[5] revealed 11 of 134 reported cases of FMH (8.2%) presented with hydrops fetalis.

Ballantyne first described hydrops fetalis in 1892 and it means "universal oedema of the newborn". It is caused by 3 main conditions: anaemia, hypoproteinaemia and cardiac failure. But in some complex cases, there may be more than one condition causing the hydrops fetalis. Hydrops fetalis can be easily diagnosed on routine ultrasound scan which will show fetal oedema, cardiomegaly, increased nuchal thickness (NT), thickened placenta and pleural or peritoneal effusion. Nonetheless, it is uncommon to have late onset hydrops which was undetected despite regular antenatal follow-up.

Case Report

A 34-year-old woman with 3 previous vaginal deliveries had an early booking at 13 weeks for her antenatal care. Routine antenatal investigations were normal. In particular, there was neither evidence of thalassaemia (mean corpuscular volume 95.4 FL) nor infection. The fetal anomaly scan at 22 weeks was normal with a nuchal fold of 4 mm. She was referred to our hospital at 31 weeks of gestation. Ultrasound scan performed at 35 weeks showed normal biometry and structure. There was no evidence of hydrops. She then presented with decreased fetal movements for several days at 38 weeks. The cardiotocography (CTG) showed decreased baseline variability and late decelerations (Fig. 1). The cervical os was 3 cm dilated. An emergency Caesarean section was performed. At delivery, the placenta was noted to be large. There was no evidence of retroplacental clots.

Upon delivery, the male neonate appeared pale and floppy at birth. The Apgar scores were 1 at 1 minute (heart rate <100 beats per minute with no spontaneous activity or breathing), 4 at 5 minutes and 7 at 10 minutes. He was resuscitated by the neonatologists and intubated. Cord

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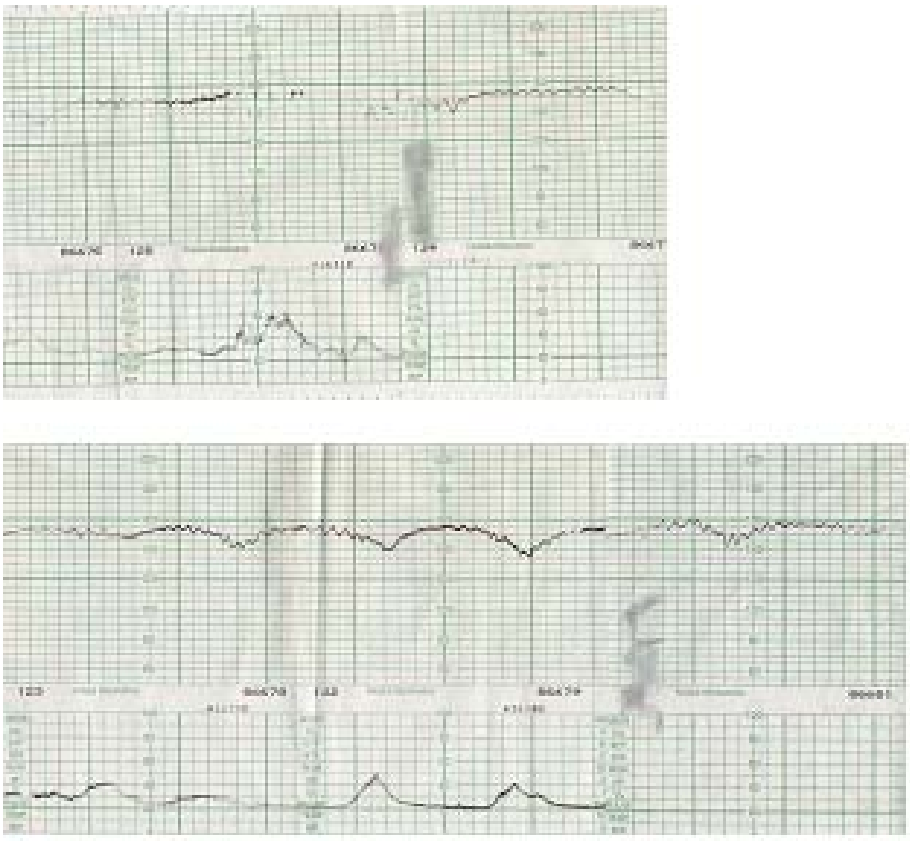

Fig. 1. CTG showing decreased variability and late deceleration.

gases confirmed severe metabolic acidosis: the arterial blood's pH, HCO_3^- and base deficit were 6.9, 10.0 mmol/L and -20.8 mmol/L, respectively, while the venous blood parameters were 7.1, 14.6 mmol/L and -12.2 mmol/L, respectively.

The hydropic neonate had a distended abdomen with ascites and an enlarged palpable liver consistent with signs of cardiac failure. An umbilical venous line was inserted for the administration of medication and resuscitation. A blood count showed a haemoglobin (Hb) level of 3.9 g/dL, haematocrit of 11.0%, reticulocyte of 20.9% and normal counts of white cells and platelets. The result was consistent with bone marrow activation caused by chronic anaemia. He required blood transfusions and inotropic support in the neonatal intensive care unit (NICU). The post-transfusion Hb was 7.9 g/dL at 3 hours of life and 10.9 g/dL at 12 hours of life.

The neonate later developed persistent pulmonary hypertension of newborn (PPHN), requiring treatment with magnesium sulphate, tolazoline and subsequently high frequency oscillatory ventilation (HFOV). Stage 2 hypoxic ischaemic encephalopathy (HIE) was also diagnosed and he was treated with intravenous phenobarbitone. The cranial ultrasound at day 1 of life showed a bulky choroid plexus on the left side. There was no evidence of periventricular leukomalacia. Follow-up scans on day 8 confirmed a grade II intraventricular haemorrhage (IVH) on the left side. On day 25, mild to moderate ventriculomegaly was noted with resolution of the IVH.

Other medical problems that developed during the NICU stay included hypoglycaemia, gentamicin toxicity, pulmonary haemorrhage, gastrointestinal tract bleeding and presumed sepsis. These were, however, transient problems that were managed conservatively.

The placental histology revealed a bulky placenta as well as an enlarged immature pattern of villi with decreased syncytial knots and increased vessels consistent with hydrops. The maternal and neonatal intrauterine infection screen was negative (including Parvovirus B19). The Kleihauer-Betke test was positive with an estimated 170 mL of fetal blood in maternal circulation, confirming the presence of massive FMH as the cause of the hydrops. The neonate was hospitalised in the NICU for 22 days and discharged at 30 days of life. During follow-up at 8 months, the child was noted to have features of spastic cerebral palsy.

Discussion

CTG was introduced over 3 decades ago and is commonly used despite the absence of evidence to support its use. The use of CTG at admission[6,7] and in the intrapartum[8] periods

for low-risk fetuses did not result any improvement in neonatal outcome. Nonetheless, CTG monitoring for the high-risk population is accepted in clinical practice as a standard of care. While this CTG is non-reassuring and predictive of poor outcome which may correlate with hypoxia of the fetal central nervous system, it does not, however indicate irreversible and severe brain damage with certainty.

Ultrasound examination was not performed at admission. Performing ultrasound in such a case is not useful as it is highly unlikely to be useful in differentiating those who had developed severe consequences from those who had not. While it would have allowed detection of hydrops, it is unlikely to have altered the management. Therefore, in our opinion, delivery by caesarean section was the correct management.

This is an unusual case of late onset non-immune hydrops secondary to FMH, resulting in cerebral palsy (CP). The prevalence of CP is estimated to be 2 to 2.5 in every 1000 live births.[9] CP from FMH is rare, based on a review of the literature. Two papers reported neurological complication from FMH. One was cerebral accident with axial hypotonia[10] and the other was asymmetric double hemiplegia with severe developmental delay.[11]

FMH can begin anytime from the mid-first trimester onwards. It is presumed to result from a breach in the integrity of the placental circulation. As the pregnancy continues, more and more women will show evidence of fetal red cells in their circulation. By term, about 50% will have detectable fetal cells. However, 96% to 98% of pregnancies have very small leaks of 2 mL[12] while massive FMH (>150 mL) occurs in 0.12% to 0.5% of pregnancies.[11] Associated causes include direct trauma to the abdomen, motor vehicle accident, abruptio placenta, vasa previa with membranous insertion, chorioangioma or choriocarcinoma, thrombus in umbilical vein, amniocentesis and external cephalic version. Nonetheless, the cause of FMH is unknown in 82% of cases.[5] In most spontaneous FMH cases, microscopic areas of placental capillary damage may result from third-trimester uterine activity.[2] Kleihauer-Betke is a reliable test for diagnosing FMH especially in the third trimester.[13] It should be performed in cases with associated risk factors such as maternal trauma and intrauterine manipulation. Pregnancies complicated by FMH often develop normally though signs of fetal decompensation may appear if the degree of FMH is high. Decreased or absent fetal movement, detection of abnormal fetal heart pattern, or hydrops fetalis are associated with severe FMH. Perinatal mortality due to massive FMH occurs in approximately 1 in 1000 deliveries.[14] The fact that FMH can occur without antecedent risk factors underscores the importance of further research, and a high index of suspicion.

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