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Survival of an infant with massive fetomaternal hemorrhage with a neonatal hemoglobin concentration of 1.2 g/dL without evident neurodevelopmental sequelae

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Abstract

Fetomaternal hemorrhage is referred to as the passage of fetal blood into the maternal circulation. Massive hemorrhage can cause severe fetal anemia, affecting fetal and neonatal outcomes. A neonatal hemoglobin concentration (Hb), which is reportedly a significant prognostic factor, of <5.0 g/dL has been reported to carry a high risk of poor outcomes (death and major morbidity). We present a case of massive fetomaternal hemorrhage with the lowest value of neonatal Hb ever previously reported in a survivor, who subsequently met all the developmental milestones at the corrected age of 18 months. A male infant born at 27 weeks gestation, weighing 998 g, presented with severe anemia with an Hb of 1.2 g/dL and an HbF level in the mother's blood of 2.4%, which led to a diagnosis of fetomaternal hemorrhage. Since there were no findings of hypovolemia, exchange transfusion was performed for prompt correction of the severe anemia without precipitating volume overload. The present case suggested that exchange transfusion might promptly correct anemia in patients with fetomaternal hemorrhage without hypovolemia without causing volume overload.

Keywords

Fetomaternal transfusion, fetomaternal hemorrhage, exchange transfusion

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Introduction

Fetomaternal hemorrhage (FMH) refers to the entry of fetal blood into the maternal circulation before or during delivery.[1] FMH in small amounts occurs frequently, with less than 0.025 mL of fetal red cells present in 74% of postpartum conditions, and it is usually without any maternal or fetal signs.[2] Using a common cutoff value of 30 mL for the diagnosis of FMH, incidence of FMH has been estimated at 3 in 1000 pregnancies.[2,3] Massive hemorrhage might have devastating effects on the fetus, such as anemia, stillbirth, and neonatal death.[1,3,4] Neonatal hemoglobin concentration (Hb) serves as one of the prognostic factors in such cases.[5–7] A neonatal Hb value of 5.0 g/dL poses high risks of death and major morbidities.[6] We present a case of massive FMH with the lowest ever reported value of neonatal Hb (1.2 g/dL) as compared to previous reports, who has met all the developmental milestones of his corrected age of 18 months.

Case

A 23-year-old primigravida at 27 + 1 weeks gestation presented to her obstetrician with decreased fetal movements. Her pregnancy had been uneventful without any risk of FMH, such as abdominal trauma, preeclampsia, amniocentesis, or abruption. Since cardiotocography showed a sinusoidal pattern, emergency cesarean section was performed. The male neonate with a birth weight of 998 g was pale, with a heart rate of more than 100 beats/min, but without spontaneous respiration. He was immediately intubated and mechanically

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ventilated. Apgar scores were 2 and 2 at 1 and 5 min, respectively. Umbilical cord pH and base deficit were 7.14 and 10.3, respectively. His laboratory blood tests indicated a hemoglobin concentration of 1.2 g/dL, hematocrit of 4.5%, and reticulocyte count of 25.9%. There was no visible bleeding or internal hemorrhage on abdominal/head ultrasonography. Both the mother's and the neonate's blood type was group A, RhD+, and the mother's antibody screen was negative. There were no findings of congenital infection in the laboratory test. The histopathological examination of the placenta showed no indications of placental tumors or inflammation. The performance of the Kleihauer et al.[8] test on the mother's blood revealed the presence of 2.4% of fetal Hb (HbF), which was estimated as 986 mL of fetal blood loss[1] (maternal hematocrit of 37%, newborn hematocrit of 4.5%, maternal HbF of 2.4%, assumed maternal blood volume of 5 L).

The neonate's mean arterial blood pressure was 26 mmHg and heart rate was 162 bpm. Chest X-ray showed a cardiothoracic ratio (CTR) of 52% and echocardiogram showed a normal left ventricular end-diastolic dimension of 12.9 mm with normal fractional shortening of 25%, and no collapse of the inferior vena cava, all of which revealed the absence of hypovolemia. Therefore, we selected exchange transfusion for prompt correction of the severe anemia without causing volume overload. While waiting for the exchange transfusion, 16 mL of red blood cells was transfused over 2 h. Within 6 h after admission, after the exchange transfusion (total amount of 180 mL), the neonates Hb level had increased to 8.9 g/dL without any signs of volume overload. After successful weaning off mechanical ventilation and extubation on day 1, his clinical course remained stable. A magnetic resonance imaging (MRI) of Brain performed at 39 weeks of corrected age showed no findings suggestive of hypoxic ischemic brain injury, such as cystic changes or abnormal intensity in the white matter. Follow-up showed that the infant has met all the developmental milestones of his corrected age of 18 months.

Discussion

The present case suggests two clinical implications. The first is that it is possible for neonates with massive FMH with a neonatal Hb of as low as 1.2 g/dL to survive without evident neurodevelopmental sequelae. The Hb in the present patient was the lowest ever previously reported in survivors of FMH. The severity of FMH depends on the hemorrhagic volume and the rate of blood loss.[4] Rubod et al.[3] reported that when blood loss rate exceeds 40 mL/kg, the patients are at a high risk for stillbirth or severe morbidity, and when it exceeds 80 mL/kg, poor patient outcomes are inevitable. Christensen et al.[6] reported that patients with neonatal Hb levels below 5.0 g/dL have a significantly higher risk of poor outcomes (death, intraventricular hemorrhage, periventricular leukomalacia, hypoxic ischemic encephalopathy, bronchopulmonary dysplasia) than patients with neonatal Hb levels above 5.0 g/dL; the rate of poor outcomes in patients with neonatal Hb below 5.0 g/dL was as high as 71%. The estimated fetal blood loss in the present case was 986 mL/kg, which was much more than the reported high-risk blood loss value of 80 mL/kg assigned by Rubod et al. for inevitable poor outcomes. Furthermore, the neonatal Hb level in the present case was 1.2 g/dL, which is much lower than the 5.0 g/dL threshold of poor outcomes reported by Christensen et al.

One of the reasons for the favorable outcome in the present case might be that the patient did not develop hypovolemia, indicating that the blood loss was chronic. In ovine experiments, removal of 30% of fetal blood over 2 h was tolerated. However, 30% of fetal sheep died after removal of the same amount of fetal blood over 10 min.[9] In another sheep fetus experiment, when the chronic bleeding persisted, the circulating blood volume was restored within 3 h because of redistribution of protein and fluid from the interstitial space. Hence, circulatory collapse was prevented.[10] In the present case, the cardiovascular findings (vital signs, CTR on chest X-ray, and echocardiogram findings) suggested that the intravascular volume was restored by compensatory mechanisms, thereby preventing circulatory collapse.

The second implication of the present case is the usefulness of exchange transfusion for FMH without hypovolemia. Although a few reports have described exchange transfusion as the treatment for severe anemia due to FMH,[11,12] only one report suggested the benefit of isovolumetric partial exchange transfusion for normovolemic FMH.[11] The present case also suggested that exchange transfusion has the potential to promptly normalize Hb levels in normovolemic FMH patients without causing volume overload, thus preventing the occurrence of intraventricular hemorrhage.[13]

Conclusion

In cases of FMH with severe anemia, it has been suggested that treatment should follow a tailor-made approach based on the patient's cardiovascular status, which should be estimated by parameters such as vital signs, chest X-ray, and echocardiogram findings, so that simple rapid transfusion is selected for cases of FMH with hypovolemia, and exchange transfusion is selected for cases of FMH without hypovolemia.

Declaration of conflicting interests

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Ethical approval

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Informed consent

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