INTRODUCTION

Alloimmune hemolytic disease of the newborn (HDN) is induced by the destruction of red blood cells (RBCs) of the neonate or fetus by maternal IgG antibodies. Maternal red cell allo-immunization occurs when the fetus is positive for an antigen that is absent on maternal red cells. The mother is stimulated to create immunoglobulin G antibodies against the positive fetal red cells which pass through the placenta.

Here, we report a case of a patient who had received packed red cell transfusion at postnatal 38 days of life due to severe Rh c and E incompatibility leading to hemolytic anemia.

CASE REPORT

The patient was born at 34 weeks of gestation by caesarean section to a 35-year healthy mother with B + blood group. She was the second child of a non-consanguineous parent. Neither parents nor siblings had a history of jaundice or anemia or gallstones. Birth weight was 2310 g, length was 42 cm, occipitofrontal circumference was 33 cm, and Apgar scores were 9 and 10 at 1 and 5 minutes, respectively.

The patient appeared normal, and fed well. When she was 5 days of age, serum bilirubin of 18.3 mg/dL was obtained as part of total serum bilirubin screening programme. This result was determined as well above the 95th percentile hour specific monogram. The mother and the patient were grouped B and B, respectively; both being positive for RhD antigen. The baby's blood group type was C+, c+, E+, e+, K-, while her mother's blood group type was C+, c-, E-, e+, K-. Our patient was diagnosed as Rh c and E incompatibility, leading to the hemolytic anemia. Minor blood group incompatibility should be considered in infants with prolonged jaundice and severe anemia, leading to heart failure.

Key Words: Hemolytic anemia. Heart failure. Anti-C. Anti-E. Jaundice. Minor blood group incompatibility.

ABSTRACT

Many different blood group systems, such as Rh, ABO, Kell, Kidd, Duffy, MNS, have been reported as causes of hemolytic disease of the newborn. Hemolysis due to minor blood group incompatibility in the fetus or newborn has been determined in isolated case reports. Here, we report a case of a patient who had received red cell transfusion due to severe Rh c and E incompatibility, leading to hemolytic anemia with heart failure. The mother and the baby were grouped B and B, respectively, both being positive for RhD antigen. The baby's blood group type was C+, c+, E+, e+, K-.

Our patient was diagnosed as Rh c and E incompatibility, leading to the hemolytic anemia. Minor blood group incompatibility should be considered in infants with prolonged jaundice and severe anemia, leading to heart failure.

Key Words: Hemolytic anemia. Heart failure. Anti-C. Anti-E. Jaundice. Minor blood group incompatibility.

INTRODUCTION

Alloimmune hemolytic disease of the newborn (HDN) is induced by the destruction of red blood cells (RBCs) of the neonate or fetus by maternal IgG antibodies. Maternal red cell allo-immunization occurs when the fetus is positive for an antigen that is absent on maternal red cells. The mother is stimulated to create immunoglobulin G antibodies against the positive fetal red cells which pass through the placenta.

Here, we report a case of a patient who had received packed red cell transfusion at postnatal 38 days of life due to severe Rh c and E incompatibility leading to hemolytic anemia.

CASE REPORT

The patient was born at 34 weeks of gestation by caesarean section to a 35-year healthy mother with B + blood group. She was the second child of a non-consanguineous parent. Neither parents nor siblings had a history of jaundice or anemia or gallstones. Birth weight was 2310 g, length was 42 cm, occipitofrontal circumference was 33 cm, and Apgar scores were 9 and 10 at 1 and 5 minutes, respectively.

The patient appeared normal, and fed well. When she was 5 days of age, serum bilirubin of 18.3 mg/dL was obtained as part of total serum bilirubin screening programme. This result was determined as well above the 95th percentile hour specific monogram. The mother and the patient were grouped B and B, respectively; both being positive for RhD antigen. The patient's blood type was B (+) and the direct antiglobulin test on the neonate's red cells was negative. Phototherapy was started by using a Giraffe Spot PT Lite (Ohmeda Medical, Laurel, MD) with a measured irradiance of 30 $\mu W/cm^2/nm$ at 40 cm from the skin. She continued to feed well and was discharged from the hospital at 6 days of age with serum bilirubin level of 9 mg/dL.

Her follow-up visit showed normal developmental milestones with appropriate increase in body weight on 8th, 15th and 30th day of life. By day 38, the patient was admitted to the outpatient clinic with jaundice and feeding intolerance. Her physical examination revealed weight 3610 g, pulse rate 168/minute, respiratory rate 65/minute, blood pressure 65/45 mm/Hg, and temperature 36.5°C. The physical examination was unremarkable except profound jaundice, decreased capillary refill and hepatomegaly 4 cm below the costal margin.

Laboratory investigation revealed total serum bilirubin 14.8 mg/dL, conjugated bilirubin 1.2 mg/dL, hemoglobin 6.3 g/dL, hematocrit 18.9%, mean cell volume 98.8fL, platelet count 220000/mm3, and reticulocyte 8.37%. Peripheral blood film showed signs of hemolysis such as anisocytosis, polychromasia, nucleated RBCs, and red cell fragments.

Detailed laboratory study was performed for differential diagnosis of the hemolysis. Blood glucose, serum electrolytes, liver enzymes, C-reactive protein, interleukin-6, urine and blood cultures were reported as being normal. G6PD screening was negative. TORCH was negative. Echocardiography was normal with minimally effected left ventricular function. When we evaluated for the non-D Rhesus blood group type, the baby's blood group type was C+, c+, E+, e+, K-.

and the mother's blood group type was C+, c-, E-, e+, K-.
She was diagnosed as Rh c and E incompatibility leading to the hemolytic anemia with heart failure. The patient was managed with red blood cell transfusion at a dose of 20 cc/kg over 4-hour. The patient was discharged on the 39th day of the life with good condition.

DISCUSSION

Although Rh c and E incompatibility is a well-defined cause of hemolytic anemia; no reports of patients with severe anemia leading to heart failure, secondary to Rh c and E incompatibility, were found in the literature. Alloimmune HDN primarily includes the major blood groups of Rhesus (Rh), A, B, AB, and O. However, minor blood group incompatibilities can also lead to significant disease. Individuals are classified as Rh negative or positive based upon the expression of the major D antigen on the erythrocyte. The original definition of alloimmune HDN was owing to Rh (D) incompatibility, which is associated with the most severe form of the disease. Other red cell allo-antibodies such as anti-c, anti-C, anti-E, and anti-e of the Rh blood group system and anti-K of the Kell blood group system have been reported occasionally as rare causes of HDN.2,3 Here, the case was of HDN due to anti-E and anti-c with evidence of hemolysis seen on blood picture (anisocytosis, polychromasia, nucleated RBCs and red cell fragments).

Minor blood group antibodies occur in response to exposure to foreign RBC minor group antigens from a previous pregnancy or transfusion. The clinical manifestations of alloimmune HDN range from mild hemolytic anemia to severe life-threatening anemia. Late onset anemia presenting 1 - 3 weeks after delivery may be observed in neonates with ABO, minor blood groups and Rh incompatibilities.4-6 Late-onset anemia may be owing to immune destruction of erythroid progenitors and/or suppression of erythropoiesis.5-7 Hemolysis due to minor blood group incompatibility in the fetus or newborn has been determined in isolated case reports. Postnatal management for involved infants is focused on treating the anemia and hyperbilirubinemia produced by hemolysis of neonatal RBCs.

The diagnosis of heart failure in children is based on a combination of characteristic signs and physical findings of impaired cardiac output, respiratory distress, and poor growth. Imaging studies generally are used to confirm the diagnosis of heart failure when there is a clinical uncertainty. We detected jaundice, feeding intolerance, tachypnea, tachycardia, hepatomegaly and delayed capillary refill in this patient. Echocardiography was almost normal with minimally affected left ventricular function.

In conclusion, minor blood group incompatibility should be considered in infants with prolonged jaundice and severe anemia leading to heart failure. If a non-pregnant woman is found to have an anti-RBC antibody, she should be counselled regarding the potential effects of the antibody on a future pregnancy.

REFERENCES