Anti Rh 17 in a multigravida with Rh-D-/•D• genotype resulting in haemolytic disease of the newborn

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Introduction

The Rh complexes -D- [1] and •D• [2] are rare. The genotype -D- is found most commonly in the offspring of consanguineous couples. People with -D- can develop anti Rh 17 when sensitised by pregnancy or transfusion which can lead to severe problems of haemolytic disease of the newborn (HDN).

Case report

A 37-year-old woman in her third pregnancy, the previous two babies were delivered by Caesarean section for cephalopelvic disproportion. There was no past history of blood transfusion.

Blood samples sent at 28 weeks gestation showed her to have blood type A Rh(D) positive. Further Rh phenotyping showed a lack of Rh antigens C, E, c and e. Her serum was found to contain an atypical antibody reacting with all Rh +ve panel red blood cells at 37 °C by the one-stage papain method, and by saline at room temperature. Anti Rh 17 was suspected in view of her Rh phenotyping, which was confirmed by reaction with all red cells except -D- and •D•.

The anti D equivalent activity of her antibody rose from 46 i.u./ml at 28 weeks to 79 i.u./ml at 32 weeks' gestation when measured by the Technicon autoanalyser machine.

Ultrasound at 29 weeks' gestation showed a disproportionate increase in fetal abdominal girth suggesting the development of early fetal hydrops.

Cordocentesis was performed at 30 and 32 weeks' gestation, which showed the fetus to have blood type O with expression of the Rh C, D, c and e antigens. The fetal red cells reacted with the mother's serum at 18 °C in saline confirming an Rh 17 positive status.

On both occasions the fetus underwent intrauterine transfusion of 80 mls of maternal blood. The pre-transfusion haemoglobin was 5.9 and 4.6 g/dl and post-transfusion 11.8 and 11.2 g/dl, respectively. The baby was delivered by Caesarean section at 33 weeks' gestation with an Apgar score of 9 at 1 min and 10 at 5 min, and weighing 2.35 Kg. The anti D equivalent activity in the mother's serum was 2020 i.u./ml and the cord blood sample had an activity of 250 i.u./ml.

Only maternal (Group A Rh-D-/•D•) red cells were detected in the neonate; the direct antiglobulin test (DAGT) was therefore negative. The cord bilirubin level was 334 µmols/l with a haemoglobin of 11 g/dl. Blood transfusion with maternal red cells and phototherapy were instigated. At four and six sweeks of age the baby was transfused with one frozen unit of Group O Rh-D- and one maternal unit respectively, the haemoglobin having fallen to 7.7 and 9.5 g/dl. Sequential bilirubin measurements showed a rapid decline to 54 µmols/l at six days' post delivery and 20 µmols/l by two weeks.

The appearance of naturally occurring anti A in the baby by four months of age precluded further transfusion with maternal red cells. Alternative treatment with erythropoietin was commenced to stimulate the baby's erythropoiesis. Five injections of erythropoietin (two of 200 i.u. and three of 400 i.u.) alleviated the requirement for further blood transfusions, with a nadir Hb of 7.5 g/dl rising to 11.6 g/dl. By ten months of age the baby was stable and did not require further therapy.

Discussion

This case illustrates several interesting features of this rare Rh phenotype. Lack of consanguinity is unusual when this Rh phenotype is discovered but the mother denied such a relationship. The development of anti Rh 17 and its association with HDN is well recognised. Serologically this antibody shows the phenomenon of agglutination of all red cells in the saline at room temperature except against other -D- cells. The quantification of the number of D antigen sites and the presence of the Evans (Rh 37) antigen [3] are not compatible with a homozygous -D- genotype suggesting the mother is probably a compound heterozygote -D-/•D•.

The use of maternal autologous donations is the most convenient way of providing compatible blood for the fetus and neonate and is commonly used when dealing with rare blood group antigens and a limited availability of cryopreserved homologous red cell units. At the time this patient first presented there were six frozen units of -D-/•D• in the U.K. and nine units cryopreserved in world blood banks that could have been utilised.

The ABO incompatibility of mother and baby led to problems when the baby developed naturally occurring antibodies, which was fortuitously overcome by the use of erythropoietin.

The mother has been advised to donate further units for cryopreservation and the risks of further pregnancies and HDN have been discussed.

References

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