



The Bombay Blood Group: How Rare is It? A Case Report and a Review of the Literature

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Authors' contributions

This work was carried out in collaboration among all authors. Author SP conceived the idea about writing the case report. Author AS was part of the management team. Author EE wrote and proofread the final version with Author SP. All authors read and approved the final manuscript.

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Case Report

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ABSTRACT

The Bombay blood group is a rare type of blood group which is very distinct from the ABO system and was first discovered in India about five decades ago. This blood group describes individuals who lack the H antigen and thus present with the anti H antibodies in addition to anti A and anti B antibodies. Bombay blood group is sometimes mistaken for blood group O. The significance of this blood group is in its ability to potentially cause fatal blood transfusion reaction and haemolytic disease of the foetus and the neonate.

Aim: This case report aims to elucidate the rare occurrence of the Bombay blood group.

Methods: We describe a case report of the pregnancy, labour and delivery of a multiparous Indian woman with the Bombay blood group in her second pregnancy.

Results: The index case had an uneventful pregnancy, labour and delivery without the need for blood transfusion. The neonate was Rhesus D positive and did not have any complications.

Conclusion: Bombay blood group is a rare blood group which can lead to blood transfusion reactions and haemolytic disease of the foetus and newborn. Pregnancy, labour and delivery should be managed in a unit with the availability of anti H blood to avoid foetal and maternal complications.

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1. INTRODUCTION

The Bombay blood group is one of the oldest and unique blood groups that has ever existed [1]. It was first discovered and described in India by Dr Bhende and colleagues about five decades ago [2]. It is a rare blood group that lacks the unique antigen known as the H antigen or the OH group on the surface of erythrocytes of individuals. Its occurrence is estimated to be about 1 in 7600 individuals in the general population, 1 in 10,000 and 1:100, 000 in the Indian community and Europe, respectively [1,3–5].

The clinical relevance of the Bombay or the OH phenotype lies in the ability to cause haemolytic disease of the foetus and the newborn (HDFN) with its attendant consequences as the antibodies cross the placenta into the foetal circulation [1,6,7]. It is unclear, but has been noted that the OH phenotype can result in blood transfusion reaction if individuals with the Bombay blood group receive blood from other groups [3,8–10]. It may be very challenging in emergency circumstances where there could be a delay in finding the Bombay blood group phenotype, and in such scenario, it is advised to only administer plasma expanders [3,10].

Patients with Bombay blood group should not be transfused with blood from other groups even in emergencies, especially O blood group, as this can lead to catastrophic haemolytic reactions as this blood is said to have a more significant amount of the H antigens [3,6]. Furthermore, the presence of the H/H antibodies can lead to alloimmunization and neonatal jaundice [6].

2. PRESENTATION OF THE CASE

The case presented is a case report of a primiparous Indian woman in her second pregnancy who was found to have the Bombay Blood group.

The Index pregnancy was registered for antenatal care at eight weeks' gestation, and there were no problems. Booking investigations showed a haemoglobin level of 103 g/l, and HIV, HBV and HCV were non-reactive. She declined chromosomal screening and had a normal dating scan. The first trimester was mostly uneventful, and she was seen in the antenatal clinic and

followed up every two weeks. Liaison was made with our haematology colleagues for the availability of anti H blood. She was also seen by a joint multidisciplinary clinic where issues surrounding the rare blood group were discussed and plans towards delivery were all put in place. She was also reviewed by the consultant anaesthetist and foetal medicine team. At 20 weeks' gestation, she had a normal anomaly scan and was commenced on ferrous sulphate to optimise her haemoglobin level.

At 28 weeks, she had an abnormal glucose tolerance test and was diagnosed with gestational diabetes. She was managed with dietary modifications in collaboration with the endocrinologist and dieticians.

Regular growth scans were started from 24 weeks and done every four-weeks until delivery and were all within the normal range. Middle cerebral artery Doppler ultrasounds were done every two weeks from 28 weeks till delivery, and there was no evidence of intrauterine anaemia.

At 36 weeks, growth scan and Doppler were all normal. The plan towards delivery was agreed with her, and she was scheduled for induction of labour between 37 and 38 weeks. Liaison with the haematologist was also made for the availability of anti H blood. She was not to have O-negative or O-positive blood transfused at any point in time. She was to have active management of the third stage of labour, avoiding prolonged labour and, if there were a need for a caesarean section, then cell salvage was to be made available with both haematologist and blood bank informed.

There was also active communication with haematology regarding blood as there were only two laboratories in England with stored anti-H blood and availability was made for us at the National Blood Bank in Liverpool. The obstetricians were also aware that blood could only stay for a maximum of 6 to 8 hours from thawing to delivery.

In the event of an emergency, it was advised by the haematologist that she could receive ABO group blood due to having had pre-transfusion testing. It was also advised that she could receive intravenous immunoglobulin (IVIG) and intravenous prednisolone to reduce the chances

of haemolytic reactions. At 36 weeks gestation, her haemoglobin level was 116 g/l.

At 37 weeks she had an uneventful induction of labour and progressed rapidly after 2 hours of artificial rupture of membranes to the delivery of a live female neonate in excellent condition, with an estimated blood loss of 350 ml. After delivery, the neonate did not exhibit any signs or symptoms of haemolytic disease of the foetus and newborn, and no admission to the special care baby unit or blood transfusion was required. The neonate's blood group was O rhesus D positive, and the direct agglutination test that was done with cord blood was negative.

The puerperium was uneventful, and she was subsequently discharged home 24 hours after delivery.

3. DISCUSSION

The Bombay blood group is a rare type of blood group with the possibilities of causing haemolytic disease of the foetus and newborn and blood transfusion reactions, although case reports are quite conflicting regarding this, and also because there are only a few published case reports in the literature [7,11]. With only 9 case reports till date, this further buttresses the rarity of this condition [3,7,11,12].

Its incidence has been reported to be about 1 in 7600 individuals in general, but it increases to about 1 in 10,000 in the Indian population and 1:1,000,000 in Europe [1,3–5].

The presence of the H antigen is first seen on the surface of erythrocytes as early as 5-6 weeks of intrauterine life and can occur even at birth although not wholly formed. The soluble A, B and H antibodies can also be seen in the amniotic fluid from about the 9th week of intrauterine life onwards, and the source is said to be of foetal in origin [6].

The H antigen is discrete, and it is usually the precursor of specific molecules known as the A and B molecules or antigens, which are formed by the addition of individual sugar molecules known as immunodominant terminal monosaccharides N-acetylgalactosamine and galactose [10].

In the ABO blood group system, we have different blood group types. Blood group A depicts individuals with the presence of the A

antigen and B antibodies; group B represents individuals with the presence of the B antigen and the A antibody; blood group AB occurs in individuals who possess the AB antigen with no corresponding antibody; and blood group O individuals have the A and B antibodies present on the surface of their red blood cells (RBCs) but without any corresponding antigen [3]. Although individuals with blood group O have the A and B antibodies present on their RBC surfaces, they lack the H antibody (since they harbour the H antigen), while individuals with Bombay blood group have A and B antibodies as well as H antibodies [1,3,10]. The ABO blood group and the Bombay blood group characteristics are depicted in Table 1.

Table 1. ABO blood group and Bombay blood group [3]

Blood group	Antibody	Antigen	Compatibility
A	B	A	Blood group A and O
B	A	B	Blood group B and O
AB	Nil	AB	Blood group A, B, AB and O
O	AB	Nil	Only group O
HH	ABH	Nil	HH

The Bombay blood group illustrates the exclusive group of individuals who lack the classical ABH antigen and have the commensurate antibodies in their plasma [10].

The Bombay blood group can sometimes be mistaken or mistyped for blood group O if the blood typing is not correctly and thoroughly carried out [3,10].

The genetic inheritance of the Bombay blood group or of the OH phenotype takes place on the long arm of chromosome 19 (19q13.3). The inheritance is said to be recessive, and the two genes responsible for this trait are also known as autosomal recessive genes [1–3,10].

On the surface of red blood cells is the H antigen, also recognised to be the precursor molecule for the formation of the A and B substrates or antigens [1,3,10]. The genes responsible for the formation of the H antigen are referred to as the H (FUT1) and FUT2 Secretor genes, and they are involved in the enzymatic activity of glycosyltransferase enzyme in the

production of the H antigen on the surface of erythrocytes by the addition of 1-fucose to a precursor substrate [3,10]. The FUT1 gene is solely responsible for the H antibodies produced on the surface of red blood cells, and the FUT2 gene has an additional role in the activity of the H antigen present in saliva, gastrointestinal and genitourinary secretions [13]. These two genes are differentiated by the presence or absence of the activity of the FUT2/Secretor gene [13].

The Bombay blood group phenotype is said to occur as a result of a missense genetic mutation in the activity of the FUT1 gene and sometimes may involve the deletion of individual coding sections of the FUT2 gene [14-16]. The resultant effect in the mutation is the formation of an inactive enzyme that is not able to produce the H antigen on the red cells, hence the formation or production of the H or OH antibody on the surface of these red blood cells [14-16].

Individuals with red blood cells of the Bombay blood group are only compatible with the serum of another individual who also has the H/H or the Bombay blood group [9,17]. Individuals with the Bombay blood phenotype can donate blood to others in the ABO blood group system but can only receive blood from fellow individuals with the distinct Bombay phenotype [3,6]. The Bombay anti-H antibodies are immunoglobulins of the IgM or IgG types and are capable of lysing erythrocytes [3,6,7,10].

The case presented is that of a primiparous lady who was identified with the anti-H blood group at booking having had an uneventful previous pregnancy. Her starting haemoglobin was initially low and was optimised accordingly. The pregnancy was managed accordingly with adequate multi-professional input from the obstetricians, anaesthetist, haematologist and blood bank services.

Individuals with the OH phenotype can only be transfused with Bombay blood because of the possibilities of fatal blood transfusion reactions and haemolytic disease of the foetus and the newborn if the individual is pregnant [3,6]. Due to the fact that Bombay blood is scarce to find with a limited number of donors, early planning for autologous blood donation would be beneficial in ensuring that blood is available in the event of postpartum haemorrhage. It may also be needed for exchange blood transfusion

for neonates with haemolytic disease [3,18,19]. In the index case, the pregnancy was highly uneventful and regular foetal-maternal monitoring with early detection of intrauterine anaemia was done with biweekly middle cerebral artery Doppler ultrasounds. This was imperative as evidence of intrauterine anaemia will always necessitate foetal medicine referral and intrauterine transfusion. She was also co-managed by the foetal medicine team.

In the antepartum period, it is also very vital to ensure that the patient haemoglobin is optimised and that any anaemia is corrected. At 36 weeks her haemoglobin was 116 g/l.

Foetal and maternal outcomes are usually good, and the index case had an uneventful normal delivery following induction of labour, with no postpartum haemorrhage or haemolytic disease of the foetus and newborn. Although this was not the case, in the event of postpartum haemorrhage, then Bombay specific typed blood must be sought and made available on time as it can be very catastrophic if needed in an emergency and it is not available. Also, if a caesarean section is required, intraoperative cell salvage may be of pivotal and lifesaving importance, especially if Bombay blood availability is delayed or unavailable [10]. Furthermore, it is often recommended that individuals with Bombay blood group should only receive Bombay blood [18]. However, other authors have also advised the transfusion of ABO group blood, K units if the pretransfusion testing was performed [10].

Although the outcome is usually good, isolated cases of adverse events have also been reported in case reports. Moores and colleagues, in their case report, noted that, of the three babies whose mothers had anti-H blood group, two were affected with HDFN and one was not. Of the two affected babies, one had an uneventful recovery after exchange blood transfusion, while the other died after this procedure [20].

Subsequent pregnancies should be managed by a multidisciplinary team. Autologous blood donation and freezing within the pregnancy period will be instrumental, as the blood may be needed in critical emergencies.

4. CONCLUSION

The occurrence of the Bombay blood group is an unusual phenomenon and, if not recognised on

antenatal booking investigations with a high index of suspicion, can lead to fatal foetal and maternal complications. A multidisciplinary team approach should be adopted in its management and the availability of anti-H blood is essential. Arrangements for anti-H blood should be made throughout pregnancy in order to minimise the rare consequences of blood transfusion reactions and haemolytic disease of the foetus and newborn.

CONSENT

All authors declare that written informed consent was obtained from the patient for publication of this case report.

ETHICAL APPROVAL

It is not applicable.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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