

ABO blood group incompatibility and primary tooth discoloration

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Abstract

A case report of discolored anterior primary teeth is presented. Medical history and clinical findings suggest an etiology of hemolytic anemia and jaundice secondary to ABO blood group incompatibility. There are no previous reports of tooth discoloration resulting from ABO blood group incompatibility.

Hemolytic anemia of the neonate may be caused by an immunologic incompatibility between a blood group of the mother and that of the child. Placental transmission of maternally formed antibody to the fetal antigen results in an antigen-antibody reaction causing lysis of red cells and eventually producing hyperbilirubinemia and jaundice in the newborn. The resultant increase in immature circulating red cells is called erythroblastosis fetalis, or erythroblastosis neonatorum. Although hemolytic anemia of the neonate may occur for a number of reasons, Rh incompatibility is invariably blamed for most cases of hyperbilirubinemia with jaundice. Major blood group (ABO) incompatibility between the mother and fetus, however, is another cause of hemolytic anemia of the neonate and also may cause hyperbilirubinemia and jaundice. ABO incompatibility occurs in 20 to 25% of pregnancies, but hemolytic anemia develops in only one out of ten cases, usually with milder symptoms than those of Rh incompatibility (Behrman and Kliegman 1983).

Pigmentation of the primary dentition may result from hemolytic anemia of the fetus or newborn. Serum bilirubin, a degradation product of hemoglobin, is deposited in forming dentin and enamel, and later becomes oxidized to biliverdin (Miller and Forrester 1959). Coloration of the teeth has been described by various authors as yellow, green, or blue green (Langmead 1912; Thursfield 1912; Tank 1951; Marsland and Gerrard 1953; Bevis 1956). The initial intensity of the color is thought by some to be related to the severity and length of the jaundiced condition (Miller and Forrester

1959), but others deny such a relationship (Marsland and Gerrard 1953). Most investigators agree that the color fades with time as affected teeth revert to a more normal appearance (Marsland and Gerrard 1953; Miller and Forrester 1959; Eisenberg and Bernick 1975).

This paper describes a case of discolored primary anterior teeth, the cause of which may be ABO blood group incompatibility.

Case Report

A first-born, normal term, three-year-old white male was referred by his family dentist with a tentative diagnosis of dentinogenesis imperfecta (DI). Physical evaluation revealed a healthy, well-nourished child whose growth and development were essentially normal. The child's medical history, as provided by the mother, was unremarkable except for a seven-day neonatal episode of icterus gravis. The mother was unable to recall the cause of this condition but did remember the detail of a positive finding on a Coombs' test. The child exhibited no osseous problems, and his sclerae appeared normal. The mother denied any familial history consistent with osteogenesis or dentinogenesis imperfecta. She also denied taking any antibiotics, specifically tetracycline, during her pregnancy.

Intraoral examination disclosed no unusual developmental findings except for a uniform opalescent appearance of the primary anterior teeth (Fig 1, adjacent page). The appearance was similar to that seen in cases of DI, with the color being slightly more blue-green. Crown morphology and enamel integrity appeared normal, however, and the discoloration was limited to the anterior teeth. Radiographic examination revealed no unusual findings with respect to root or pulp chamber morphology. Thus, despite the opalescent appearance of the primary maxillary and mandibular central and lateral incisors, all additional findings were inconsistent

with a diagnosis of DI. The mother stated that the tooth discoloration was much less noticeable now than it had been when the teeth were newly erupted.

Subsequent bloodwork on both the mother and child ruled out Rh factor incompatibility. Antibody in the mother against a low frequency antigen in the child (minor blood groups) was ruled out by postneutralization compatibility of maternal and child sera. The tests confirmed that the mother (Blood Group O) and child (Blood Group A) have an AO incompatibility. Later, hospital birth records were obtained which agreed with our laboratory findings and listed a final diagnosis of jaundice secondary to AO incompatibility.

Discussion

When a child presents with discolored teeth, the dentist must decide whether the enamel or dentin is affected, whether the discoloration is isolated or associated with other problems, and whether the discoloration is genetic or environmental in etiology. In the case reported here, the enamel was judged by clinical and radiographic inspection to be normal in thickness, density, and translucency. The discoloration did not match that of the only type of amelogenesis imperfecta that has significant discoloration (pigmented, hypomature amelogenesis imperfecta). Furthermore, there were no dysmorphic features to suggest a multisystem disorder as the cause of the discoloration. In particular, there was no history or evidence of fractures, joint laxity, deafness, blue sclerae, or capillary fragility to suggest a connective tissue disorder, such as osteogenesis imperfecta. Finally, the family history was negative for other isolated discolorations of the teeth. The family history also was negative for enzyme deficiencies, hemolytic disease, and anemia.

There was, however, a positive history for hemolytic disease in the newborn period as a consequence of AO blood group incompatibility. The hue of the discoloration,

its distribution, and its change with time match the discoloration caused by Rh blood group incompatibility.

There are few reports in the early literature on staining of teeth due to icterus gravis. This is probably related to the poor survival rate of affected children (Marsland and Gerrard 1953). Thursfield (1912) and Langmead (1912) were the first to propose that dental changes might be associated with jaundice or hemolytic anemia of the neonate. Each reported a separate case of yellow-green stained anterior teeth in infants who were severely jaundiced at birth. The color was reported to have started as bright yellow, changing to green, and finally fading to a more normal color. Both authors proposed that the stains were due to "bile pigments" deposited in the forming teeth during the first few weeks after birth. Since that time, others have confirmed and documented that the cause of tooth staining was deposition of bilirubin in the forming tooth structure (Losch, Brown, and Boyle 1940; Miller 1951; Tank 1951; Marsland and Gerard 1953; Bevis 1956; Miller and Forrester 1959; Rosenthal et al. 1976).

To this date, discoloration of the primary teeth due to jaundice at birth has been reported only for hemolytic anemia of the neonate associated with Rh incompatibility. The mechanism for ABO blood group incompatibility is essentially the same. ABO factors in the fetus or newborn have a low antigenicity, which may account for the low incidence of ABO-induced hemolytic anemia. Hemolytic anemia usually is observed when the mother is Blood Type O and the infant is Blood Type A or B, with A being more antigenic. Unlike Rh incompatibility, in which sensitization of the mother usually occurs at the time of birth of the first child who remains unaffected, ABO incompatibility may be seen in the first child. In AO incompatibility, antibodies to the A antigen are present in the 7S (IgG) fraction of gamma globulin, which can cross the placenta, causing an AO isoimmune hemolytic disease in the first-born infant. If the antibody levels are high enough, the infant can be affected severely (Behrman and Kliegman 1983).

Conclusion

ABO incompatibility represents a spectrum of disease that has clinical manifestations at its extreme (Desjardins 1972). Generally, jaundice within the first 24 hr after birth is the only clinical manifestation. In the case presented, all findings were normal except for jaundice at birth, a positive Coombs' test, and documented AO incompatibility between the mother and child. The distribution and pattern of dental staining in the anterior segments were similar to descriptions of dental findings of hemolytic anemia in the children with a history of Rh incompatibility (Ball 1974). Moreover,

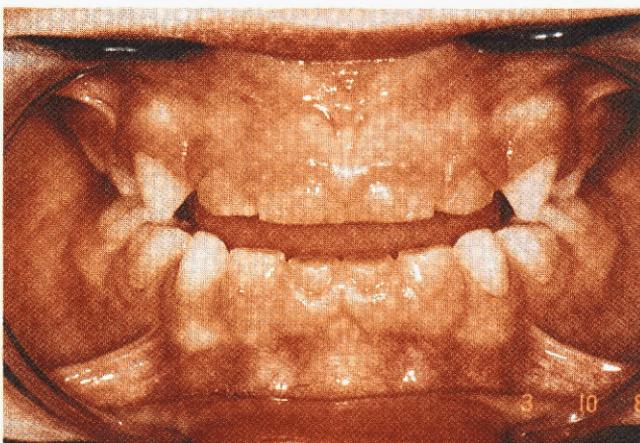


Fig 1. Appearance of dentition at three years of age.

the parents reported that the discoloration had faded with time. Thus, since all findings relating to primary tooth discoloration were negative except for hemolytic anemia of the neonate, a diagnosis of anterior primary tooth discoloration caused by hyperbilirubinemia due to AO blood group incompatibility is proposed.

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