Maternal Cautopyreiophagia as a Rare Cause of Neonatal Hemolysis: A Case Report

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Abstract

Hyperbilirubinemia in the first 24 hours of life in a newborn is pathologic, necessitating additional evaluation. We report the first case of hemolysis and subsequent hyperbilirubinemia in an otherwise normal term neonate resulting from oxidative stress in the form of maternal cautopyreiophagia: the ingestion of burnt matchstick heads. During the third trimester of pregnancy, the infant’s mother consumed more than 300 burnt matchstick heads weekly for 4 weeks. Matches contain potassium chlorate, a powerful oxidant that when ingested can ultimately lead to the destruction of erythrocytes, disseminated intravascular coagulation, kidney injury, or death. The infant’s bilirubin rose as high as 17 mg/dL at 22 hours of life; however, the infant did well with a brief course of phototherapy. This case highlights the importance of prenatal questioning about maternal ingestion of potentially oxidative substances and assessing the possible risk for the infant.

Case

We cared for a full-term African American girl delivered via cesarean delivery at 39 weeks’ gestation to a 29-year-old mother. The pregnancy was complicated by the development of pica during the third trimester. The mother reported she was ingesting >300 burnt matchsticks per week, a behavior known as cautopyreiophagia. Because she was unable to modify her behavior and because of the potential for toxicity, the obstetricians proceeded with an elective induction at term. Routine prenatal laboratories were reassuring; her blood was type A− and negative for the presence of red cell antibodies. The infant was vigorous at delivery, with Apgar scores of 8 and 9. She was appropriate for gestational age and her physical examination was normal. At 22 hours of life the infant was found to...
have an elevated screening transcutaneous bilirubin level of 17 mg/dL, high risk according to the Bhutani chart. Serum unconjugated bilirubin was confirmed at 16.7 mg/dL. Additional laboratory evaluation showed an elevated conjugated bilirubin of 1.65 mg/dL (normal, 0.1–0.3 mg/dL), aspartate transaminase of 170 U/L (normal, 5–32 U/L), and alanine transaminase of 37 U/L (normal, 4–33 U/L). Reticulocyte count and lactate dehydrogenase were also elevated at 19% (normal, 0.5%–2.2%) and 1381 U/L (normal, 135–225 U/L), respectively. The infant’s complete blood count was significant for an elevation in the number of white blood cells at 62.1 x 10^9/L (normal, 9–30 x 10^9/L), normal hematocrit of 48% (normal, 43%–65%), and anisocytosis, macrocytes, and polychromasia on peripheral blood smear. Her blood type was AB+, and direct antiglobulin testing was negative.

The infant was treated with phototherapy for 72 hours until her bilirubin level was 8.1 mg/dL, and she did not experience a rebound in bilirubin levels. She completed a 7-day course of empirical gentamicin and ampicillin; blood cultures were negative for growth. She was discharged from the hospital on day of life 8. Follow-up at 2 months of life showed 2 normal newborn screens and a normal G6PD level of 15.1 U/g hemoglobin (normal, 7–20.5 U/g). At her 9-month well child visit the infant was growing along the fourth percentile for weight, 15th percentile for height, and 13th percentile for head circumference. She was meeting age-appropriate developmental milestones and her physical examination was reassuring. Laboratory evaluation revealed a normal hematocrit of 34.3%, reticulocyte count 1.6%, total bilirubin 0.1 mg/dL, conjugated bilirubin <0.2 mg/dL, and lactate dehydrogenase 243 U/L. The patient’s transaminitis was also improving: aspartate transaminase 62 U/L and alanine transaminase 54 U/L. Hemoglobin electrophoresis showed hemoglobin A 96.8%, hemoglobin A2 2.7%, and hemoglobin F 0.5%, with a negative result for variant hemoglobins. Osmotic fragility was not elevated.

DISCUSSION

Jaundice is one of the most common problems encountered in term newborns. Hyperbilirubinemia presenting in the first 24 hours of life is considered pathologic, with causes including infection, liver dysfunction, metabolic disease, hemolytic disease secondary to blood type incompatibility, erythrocyte membranopathies, and erythrocyte enzyme deficiency such as pyruvate kinase or G6PD. The erythrocyte is reliant on the enzymes present in the hexose monophosphate shunt, such as G6PD, to prevent oxidant injury via the production of reduced nicotinamide adenine dinucleotide phosphate (NADPH). Oxidants such as the superoxide anion and hydrogen peroxide are formed in the erythrocyte via reaction of hemoglobin with oxygen and can also be produced by exogenous factors such as drugs, infection, and oxidant exposure. Within the erythrocyte, GSH is generated via a reaction between oxidized glutathione (GSSG) and NADPH, catalyzed by the enzyme glutathione reductase. The enzyme glutathione peroxidase then drives the reaction between the produced GSH and oxidant to form water and GSSG (Fig 1). Without NADPH and the glutathione pathways, oxidant exposure leads to membrane injury, methemoglobin formation, osmotic fragility, and destruction of erythrocytes.4

Ingestion of matches during pregnancy dates back decades, but literature thoroughly describing cautopyreioaphaga is scarce.5–7 In the United States, commercially available matchsticks contain sulfur, potassium...
To reach the toxic dose of 5 g of KClO₃, found in matchsticks, is a powerful oxidant that animal and in vitro human studies have shown to cause oxidative injury by reducing levels of GSH and inducing erythrocyte membrane rigidity, with subsequent lysis of erythrocytes. The human adult toxic dose of KClO₃ can be reached with the ingestion of ~400 unburned matches. Data on ingestion of burnt matchstick heads are not robust because their composition varies depending on how long the match was allowed to burn. However, reports in the forensic sciences reveal the presence of KClO₃ in both unburned and burnt matches, although no quantification is provided. In this case, the mother lit the matches and quickly extinguished them, leaving some KClO₃ intact. We hypothesize that our patient developed hemolysis, acute hepatic injury, and hyperbilirubinemia secondary to toxic levels of KClO₃ remaining in the burnt matchsticks her mother ingested. Thus, this is the first reported case of neonatal hemolysis with subsequent hyperbilirubinemia associated with maternal cautopyreiophagia. Although the infant did well clinically after the initial in utero exposure period, the long-term effects of these oxidants are not yet known. In addition, many household disinfectants, cosmetics, mouthwashes, toothpastes, and medications contain oxidative agents such as chlorates. This case highlights the importance of prenatal questioning about maternal ingestion of nonfood substances, specifically oxidants, because of the potential risk for the infant.

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