

Perinatal/Neonatal Case Presentation

Management of Severe Hyperbilirubinemia in the Newborn: Adrenal Hematoma Revisited

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A 4-day-old male infant presented with complaints of jaundice on the third day of life. He was full-term and appropriate for gestational age and born to unrelated parents. All laboratory investigation tests were normal except total serum bilirubin of 27.4 mg/dl with a direct bilirubin 0.29 mg/dl. Abdominal and cranial ultrasonography (US) was performed on sixth day of life because of severe hyperbilirubinemia. Abdominal US revealed adrenal hematoma. Enclosed hematomas may cause significant unconjugated hyperbilirubinemia in absence of other high-risk conditions.

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INTRODUCTION

In the neonatal period, unconjugated hyperbilirubinemia (UHB) is common and is associated with a variety of physiologic and pathologic conditions. Extravascular blood in particular can lead to UHB via excess bilirubin production. Common forms of blood extravasation in term infants are skin bruises and cephalic hematoma.¹ Adrenal hematoma is an unusual event in newborns, and presents with an acute onset.² Here, we present an infant whose adrenal hematoma was associated with severe hyperbilirubinemia that responded poorly to high intensity phototherapy.

CASE REPORT

A 4-day-old male infant was admitted to our hospital for jaundice. The medical history did not disclose consanguinity; his mother was

25 years old and this was her first pregnancy. In another center, the child was born with normal spontaneous vaginal delivery at 38 weeks of gestation; weighing 3370 g (75th to 90th percentile) and having no apparent complications, with Apgar scores of 9 and 10 at 1 and 5 minutes, respectively. The jaundice started on the third day after birth.

The initial hemodynamic and respiratory parameters of the infant were stable on admission. His weight was 3420 g, his length was 50 cm (50th to 75th percentile) and his occipito-frontal circumference was 34 cm (50th to 75th percentile). No external bruising or bleeding was visible. The skin and sclera were jaundiced, but the rest of the physical examination was unremarkable.

Laboratory results were as follows: total serum bilirubin: 27.4 mg/dl, direct bilirubin: 0.29 mg/dl. Hemoglobin: 15.1 g/dl, hematocrit: 45.4%, mean corpuscular volume: 103.4 fl, mean corpuscular hemoglobin concentration: 33.5 g/dl, white blood cell count: $14.7 \times 10^9/l$, platelet count: $112 \times 10^6/l$. There was no blood group incompatibility. Blood smear showed no signs of erythrocyte hemolysis. Direct antibody test (Coombs') was negative and reticulocyte count was 1.9%. Blood chemistry was as follows: urea: 24 mg/dl, creatinine: 0.3 mg/dl, Na: 137 mmol/l, K: 4.9 mmol/l, AST: 57 U/l, ALT: 24 U/l, GGT: 265 U/l, albumin: 3.4 mg/dl and the ratio of total serum bilirubin to albumin was 5.3. Urine analysis and culture were normal, and the urine tested negative for reducing substances. Serum glucose-6-phosphate dehydrogenase, pyruvate kinase and thyroid concentrations were all normal. The auditory brainstem response test was noncontributory.

After phototherapy has been initiated (neoBLUE™, Natus Medical Inc., USA), peripheral capillary levels of total bilirubin were found to be 21.3 mg/dl and 21.1 mg/dl at the 4th and 10th hours, respectively. Total bilirubin level decreased below the limit of exchange transfusion. However, due to the severity of hyperbilirubinemia, phenobarbital was administered at a dose of 5 mg/kg. Under the combined therapy of phototherapy plus phenobarbital, the total bilirubin levels at the 16th, 22nd and 34th hours were, respectively, 20.8 mg/dl, 19.2 mg/dl and 13.2 mg/dl. Throughout this process, there was no decrease in the measured capillary hematocrit level.

As the patient's severe hyperbilirubinemia suggested a likely diagnosis of internal hemorrhage, cranial and abdominal ultrasonography (US) were performed. Cranial US was normal but abdominal US revealed a 26×30 mm right adrenal hematoma (Figure 1).

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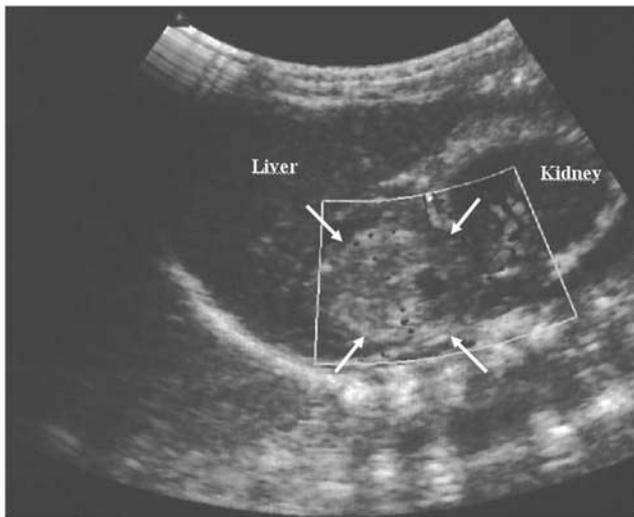


Figure 1. Transabdominal US demonstrating a heterogeneous soft-tissue mass lesion consistent with an adrenal hematoma on the right side (arrows).

We then performed adrenal function tests and blood coagulation studies both of which were normal. At follow-up after 1 month, the patient was quite normal.

DISCUSSION

The American Academy of Pediatrics (AAP) Subcommittee on hyperbilirubinemia has proposed guidelines for the management of hyperbilirubinemia in newborn infants whose gestational age is 35 weeks or more.^{3,4} In those guidelines, cephalic hematoma and significant bruising are identified as major risk factors for hyperbilirubinemia.⁴ However, there are no data pertaining to other sites where internal hematomas may occur such as spleen, liver, internal cranium and adrenal glands. Although adrenal hematoma is a relatively uncommon clinical problem in the newborn period, it might create severe hyperbilirubinemia when it occurs.

The incidence of adrenal hemorrhage ranges from 1.7 per 1000 autopsied newborn infants to approximately 3% of infants subjected to screening by abdominal US.^{5,6} It affects the right side more than the left side and is bilateral in 10 to 15% of the cases.⁷ Moreover, in newborns with severe hyperbilirubinemia, the frequency of adrenal hematoma is yet unknown. In our case, we have performed US examinations as hyperbilirubinemia persisted on the second day of phototherapy where normally intensive phototherapy is expected to produce a decrement of 30 to 40% in the initial bilirubin level after 24 hours.⁴

In conclusion, when dealing with protracted severe hyperbilirubinemia, we encourage consideration of internal hematoma as the cause. Our case of adrenal hemorrhage represents one such cause. In our opinion, the AAP management recommendations should include internal hemorrhage on the differential diagnostic list of causes for severe hyperbilirubinemia.

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