

## A rare cause of hyperbilirubinemia in a newborn: bilateral adrenal hematoma

Selahattin Katar<sup>1</sup>, Hatice Öztürkmen-Akay<sup>2</sup>, Celal Devocioğlu<sup>1</sup>, Mustafa Taşkesen<sup>1</sup>

Departments of <sup>1</sup>Pediatrics, and <sup>2</sup>Radiology, Dicle University Faculty of Medicine, Diyarbakır, Turkey

**SUMMARY:** Katar S, Öztürkmen-Akay H, Devocioğlu C, Taşkesen M. A rare cause of hyperbilirubinemia in a newborn: bilateral adrenal hematoma. Turk J Pediatr 2008; 50: 485-487.

Hyperbilirubinemia is an important health problem in newborns. The most common causes are Rh and ABO incompatibility, hemolytic anemias, enzyme deficiencies, sepsis, hypothyroidism, pyloric stenosis and breast-milk jaundice. Adrenal hemorrhage is a rare cause of hyperbilirubinemia in the neonate. We present a six-day-old newborn with hyperbilirubinemia and suprarenal hematoma who was born at home without assistance of healthcare personnel. Adrenal hematoma should also be considered in the differential diagnosis of hyperbilirubinemia, particularly in newborns that experienced a difficult delivery.

**Key words:** hyperbilirubinemia, bilateral adrenal hematoma, ultrasound, newborn.

Neonatal adrenal hemorrhage affects 0.2% of newborns and it usually develops due to birth trauma, large birth weight, hypoxia and asphyxia. However, it may also occur spontaneously<sup>1,2</sup>. Adrenal hematomas are usually unilateral and 70% of them are located on the right side. Its bilateral form is rare and represents only 10% of all cases<sup>2-4</sup>. Clinical features vary, depending on the amount of blood loss. Frequent clinical manifestations are anemia, persistent jaundice and abdominal distension associated with an abdominal mass. Here, we present a rare case of hyperbilirubinemia caused by adrenal hematoma in a newborn, which was identified by ultrasonography.

### Case Report

A six-day-old boy was admitted to our hospital with jaundice appearing two days after his birth. The patient was born at home in the 38<sup>th</sup> week of gestation (birth weight 4000 g) without any healthcare staff assistance. He was cyanotic at birth and had not cried immediately after delivery. He had been breastfed since delivery and no vaccines had been administered. Parents did not have a history of consanguineous marriage and their five children were alive and healthy.

On admission, findings were as follows: weight 3600 g, length 51 cm, head circumference 36 cm (50<sup>th</sup> percentile), axillary temperature

36.7°C, blood pressure 57/35 mmHg and heart rate 148/min. General appearance was poor, hypoactive and very icteric. He was lethargic with his head slightly dorsiflexed. Liver was palpable 2 cm below the costal margin at midclavicular line. A soft mass of 5x5 cm was palpable on the left side of the abdomen close to the localization of the kidney. Newborn reflexes were diminished.

Blood count revealed the following: Hb 15.7 g/dl, Hct 43.5%, WBC 17,100/mm<sup>3</sup>, erythrocytes 4,700,000/mm<sup>3</sup>, and platelets 503,000/mm<sup>3</sup>. On peripheral smear, 40% polymorphonuclear (PMN) leukocytes and 60% lymphocytes were found, and thrombocytes were clustered and insufficient in number. There was neither spherocytosis nor elliptocytosis. Sickling test was negative and reticulocytes constituted 1%. Osmotic fragility test was negative and glucose-6-phosphate dehydrogenase (G6PD) screening was normal. Blood chemistry findings were as follows: urea 44 mg/dl, creatinine 0.4 mg/dl, Na 139 mEq/L, K 5.3 mEq/L, ALT 34 IU/L, AST 85 IU/L, blood glucose 68 mg/dl, total bilirubin 24 mg/dl, and C-reactive protein (CRP) 0.18 mg/dl. Urine analysis findings, TORCH, T4, thyroid stimulating hormone (TSH), Tandem-mass, prothrombin time (PT) and activated partial thromboplastin time (aPTT) evaluations were normal. Both the

patient's and his mother's blood groups were A Rh (+), and direct Coombs test was negative on microscopic examination. Blood and urine cultures were negative. Abdominal ultrasound (US) examination revealed a cystic lesion with a thick wall measuring 45x24x28 mm on the right adrenal gland and a high density solid component measuring 45x42x27 on the left adrenal gland (Fig. 1). Abdominal computerized tomography (CT) detected two hypodense masses, one measuring 30x25 mm on the right adrenal gland and the other measuring 40x35 mm on the left adrenal gland (Fig. 2). Cranial ultrasonography was normal. Phototherapy was administered and after two days serum bilirubin level decreased to 8.9 mg/dl. The patient was discharged on the 13<sup>th</sup> day of the postnatal period. Ultrasonographic examination



Fig. 1. Bilateral adrenal cystic lesions with high density and solid components.

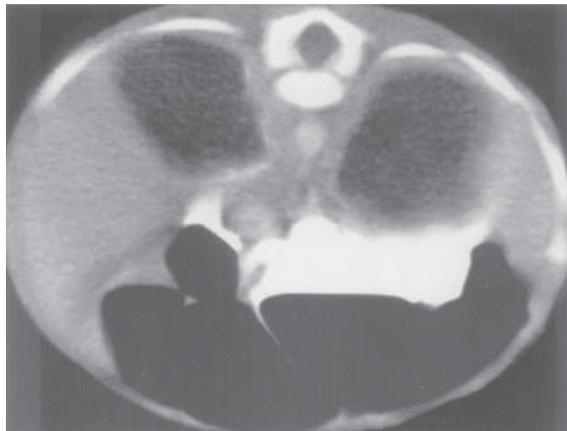


Fig. 2. Conspicuous bilateral adrenal lesions in cystic density (abdominal CT).

was done six months later, and demonstrated resolution of the adrenal hematoma (Fig. 3). At present, the patient is one year old and follow-up examinations did not reveal any residual jaundice or any sign of kernicterus.



Fig. 3. Ultrasound image of left adrenal zone, seen as almost normal.

## Discussion

Hyperbilirubinemia is an important health problem in newborns. The most common causes are Rh and ABO incompatibility, hemolytic anemias, enzyme deficiencies, sepsis, hypothyroidism, pyloric stenosis and breast-milk jaundice. However, the underlying cause of hyperbilirubinemia can not be determined in 30-40% of patients<sup>5</sup>. Although intraabdominal (liver, suprarenal) hemorrhage rarely occurs in patients born with difficult vaginal delivery, it may also be a cause of hyperbilirubinemia<sup>1</sup>. Perinatal asphyxia (in babies of diabetic mothers), trauma, coagulation difficulties and septicemia may be the cause<sup>1,6</sup>. In the present case, there was a history of difficult delivery without assistance of any healthcare staff. As bleeding diathesis was not considered to be the cause, difficult delivery was believed to be responsible for the adrenal hematoma.

Newborn adrenal hematomas often occur during the first several days following birth<sup>4,6</sup>. As our case presented with hyperbilirubinemia and abdominal mass, an abdominal US examination was performed at the 6<sup>th</sup> day and bilateral cystic lesions were found. Most of the cystic lesions originate from adrenal gland hemorrhages<sup>7</sup>. Rarely, adrenal cyst cases associated with Beckwith-Wiedemann syndrome have been reported, and the only bilateral cyst case with Beckwith-Wiedemann syndrome was reported

by Akata et al.<sup>8</sup>. No clinical features indicating Beckwith-Wiedemann syndrome were present in our case. Neuroblastoma should also be considered in the differential diagnosis of adrenal gland lesions<sup>6</sup>. Radiological appearance varies depending on the age of hematoma. While acute hematomas appear echogenic on US, they liquefy with time and assume a cystic appearance. They shrink and decrease in size and may calcify with aging. Generally, it spontaneously undergoes resorption within 4-6 weeks<sup>6,8</sup>. Thus, US follow-up is of great importance for ruling out neuroblastoma. Although there is no specific US sign for adrenal hematoma, demonstrating a decrease in lesion size and observing ECHO changes that are expected to take place in relation with the age of the lesion may suffice for the diagnosis of adrenal hematoma by US without any need for further imaging. Our patient had progressive jaundice starting from the 2nd day, and adrenal hematoma was considered to be the underlying cause of hyperbilirubinemia after ruling out other possible causes. During US follow-ups, hematomas gradually decreased in size and resolved after six months.

Adrenal hematomas are often limited with the gland capsule. Occasionally, massive adrenal hematomas are reported to break free and cause scrotal hematoma through retroperineal expansion due to the rupture of the capsule<sup>1-2,6,9</sup>. Complications such as disseminated intravascular coagulation (DIC) have been reported during the course of this disease<sup>1</sup>. In our patient, hematoma was limited with the capsule on both sides, and laboratory and clinical findings were within normal range, except for hyperbilirubinemia.

In conclusion, along with other possible causes, adrenal hematoma should also be considered in the differential diagnosis of hyperbilirubinemia of newborns, particularly in newborns that experienced a difficult delivery. US seems to be the most appropriate method for determining the etiology because it does not necessitate radiation exposure, can be easily applied and repeated, and does not require sedation.

#### REFERENCES

1. Duman N, Ören H, Gülcan H, Kumral A, Olguner M, Özkan H. Scrotal hematoma due to neonatal adrenal hemorrhage. *Pediatrics Int* 2004; 46: 360-362.
2. Vittorio M, Galluzzo M, Patti G, Mazzoni G, Calisti A, Valenti M. Scrotal hematoma due to neonatal hemorrhage: the value of ultrasonography in avoiding unnecessary surgery. *Pediatr Radiol* 1997; 27: 672-674.
3. Ausserer O, Ortore PG, Sarra A, von Fioreschy G. Adrenal hemorrhage in the newborn. *Prog Pediatr Surg* 1983; 16: 107-11.
4. Kuhn PJ, Slovis LT, Haller OJ. Caffey's Pediatric Diagnostic Imaging (10th ed.). Volume I. Philadelphia: Mosby; 2003: 245-246.
5. Sanpavat S. Exchange transfusion and its morbidity in ten-year period at King Chulalongkorn Hospital. *J Med Assoc Thai* 2005; 88: 588-592.
6. Jacobsson H, Kaiser S, Granholm T, Ringertz GH. Neonatal adrenal haemorrhage at bone scintigraphy: a case report. *Pediatr Radiol* 1998; 28: 896-898.
7. Wang LJ, Wong YC, Chen CJ, Chu SH. Imaging spectrum of adrenal pseudocysts on CT. *Eur Radiol* 2003; 13: 531-535.
8. Akata D, Haliloglu M, Ozmen MN, Akhan O. Bilateral cystic adrenal masses associated with the incomplete form of Beckwith-Wiedemann syndrome. *Pediatr Radiol* 1997; 27: 1-2.
9. Avolio L, Fusillo M, Ferrari G, Chiara A, Bragheri R. Neonatal adrenal hemorrhage manifesting as acute scrotum: timely diagnosis prevents unnecessary surgery. *Urology* 2002; 59: 601.