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Case Report – 2

Adrenal hemorrhage presenting as severe indirect hyperbilirubinemia in a neonate

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Introduction

Adrenal haemorrhage is an uncommon cause of jaundice[1]. The clinical presentation is non-specific; abdominal mass, jaundice, painful swelling of the scrotum, and rarely acute adrenal crisis are reported[1,2]. High vascularity and larger size of the adrenal glands compared to the body weight, hypoxia, coagulopathy, and difficult delivery make neonates susceptible[2].

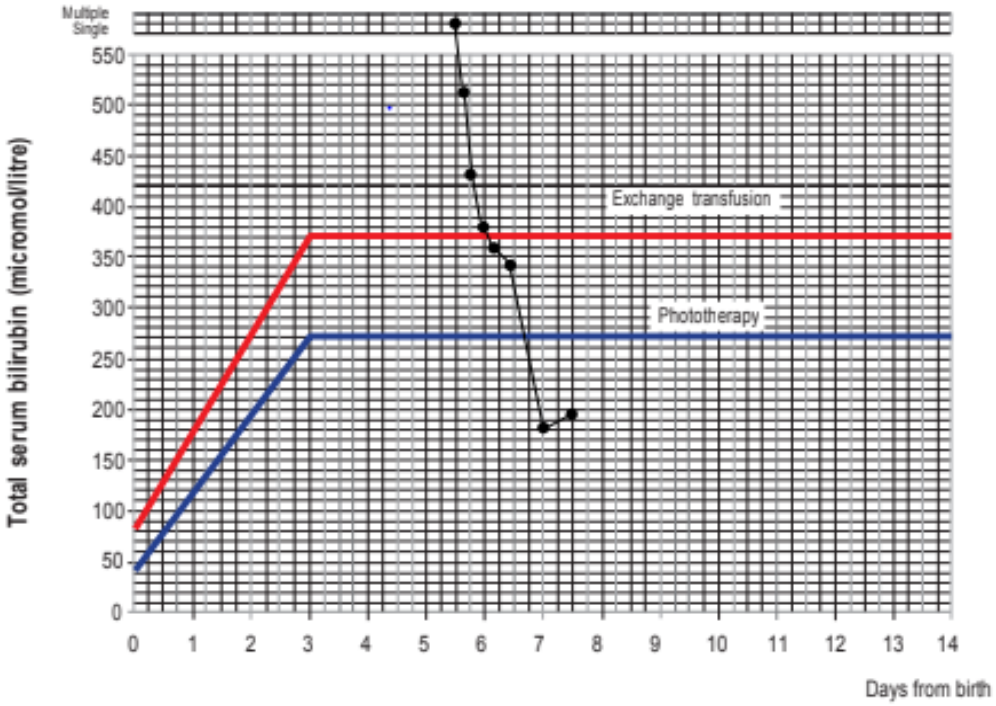
Severe indirect hyperbilirubinemia is when the level of bilirubin is >425 μmol/L[3]. We present a neonate who had a right-side adrenal hemorrhage when evaluated for jaundice.

Case history

A baby boy, the second born to healthy non-consanguineous parents at term presented to the hospital with concerns of jaundiceon the sixth day of life. Clinical examination revealed a febrile (38^0C), active, alertbaby, with jaundice up to feet. There was no cephalhaematoma. Features of bilirubin encephalopathy were not present; the baby had good sucking, normal tone, posture, and reflexes. The weight loss on the sixth day was 10% and there were no risk factors for late-onset sepsis despite the baby being febrile. The baby was delivered vaginally (birth weight – 3.28kg) in the left occiput-posterior position with difficulty, however, he was in good condition afterwards and was discharged the next day. The neonatal check-up was normal. The mother's blood group is A positive with no jaundice in the first baby.

The baby was started on phototherapy, intravenous immunoglobulins, and intravenous antibiotics pending investigations. The decline in bilirubin with phototherapy was very rapid (150 μmol/L reduction at 6 hours) that an exchange transfusion was not required, and at 12 hours of phototherapy, the bilirubin level was below the exchange transfusion level (Figure 01). The baby required phototherapy only for 48 hours.

Figure 01

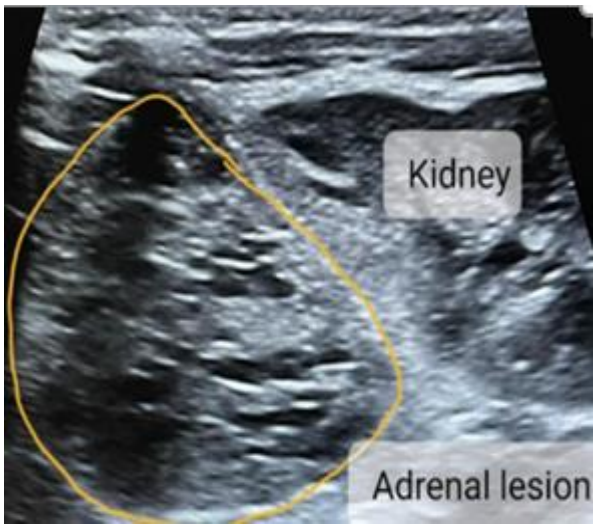


The initial investigations revealed;
Normal Full Blood Count [Hb – 15.9 g/dl (11-17), WBC – 23.36 × 10^3 /µL (13-38), N – 62% (46-74), L – 26% (13-30), Platelets – 237 × 10^3 /µL (150-350)], Marginally high C-reactive protein – 7.3 mg/L (<6), Elevated indirect bilirubin: Total serum bilirubin – 579.5 µmol/L (<270), Indirect bilirubin – 516 µmol/L, Direct bilirubin – 63.5 µmol/L: i.e. 11% of total bilirubin (<20%), No evidence of haemolysis: Reticulocyte count– 1.36% (< 5%), Normal blood picture, Blood group – O positive, Direct antibody test –negative, Normal electrolytes [Na^+- 141.5mmol/l (135-148), K^+- 5.1mmol/l (3.5-5.1)], Normal renal function [Serum creatinine – 66.2µmol/l (14-86)], Normal liver enzymes [AST – 94 U/l (26-98), ALT – 33.4 U/l (6-60)].

The immunoglobulins were discontinued after 4 hours as there was no definitive evidence of antibody-mediated haemolysis. The antibiotics were stopped at 48 hours as the blood culture was negative.

Since this was a severe indirect hyperbilirubinemia an ultrasound scan (USS) - abdomen was performed to look at the liver architecture and to identify any intrabdominal hematomas. This revealed a mass in the right-side supra-renal area suspicious of aadrenal hemorrhage, an incidental finding (Figure 02). However, the cardiovascular status of the baby was normal throughout, with good urine output, and there was no hypoglycemia or hyperkalemia.

Figure 02



There is a 3.5 cm × 3.1 cm × 3.8 cm size well defined lesion in the right supra-renal region with multiple cystic spaces. It does not cross the midline but bulges into the upper pole of the right kidney. There is no significant vascularity or calcifications. The left adrenal gland is normal

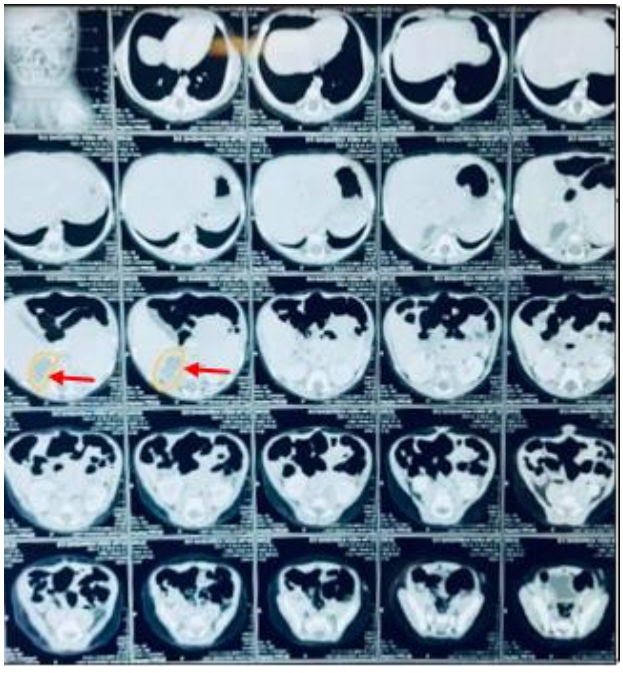
Normal liver architecture, No intrabdominal haematomas, No para-aortic lymphadenopathy

Kidneys, ureters, and bladder are normal

Possible organizing adrenal haemorrhage, however, neuroblastoma cannot be excluded

Since a repeat USS-abdomen at three weeks of age showed minimal interval resolution of the right adrenal mass a CECT – chest, and abdomen were performed to exclude neuroblastoma (Figure 03). The 24-hour urinary Vanillyl mandelic Acid (VMA) levels were normal [0.2 mg/24 hours – normal range (1 – 11)]. At three-month review, the baby is doing well and the USS-abdomen showed thatthe right adrenalmassis resolving.

Figure 03



The lung fields appear normal, there is no hilar or mediastinal lymphadenopathy, there are no mediastinal masses

There is a cystic lesion in the right suprarenal area measuring 2.3cm × 1.9cm with a thin non-enhancing hyperdense wall, the right adrenal gland cannot be identified separately

There is no para-aortic lymphadenopathy, the liver, kidney, ureters, left adrenal gland and bladder are normal

Right adrenal cyst with early features of wall calcifications, no evidence of distant metastasis, **the appearance favors a haemorrhagic cyst**

Discussion

Neonatal adrenal hemorrhage occurs more commonly in the right side (70%) as in our case[4]. The highly vascular adrenal glands are sensitive to changes in the venous pressures precipitated by trauma in difficult deliveries[2]; the occiput-posterior position is a risk factor for prolong second stage of labor which was present in this case[5]. The clinical severity of the adrenal haemorrhage will depend on its extent and the compression of the gland; a large haemorrhage will compromise the gland easily and cause severe jaundice. Hence, when babies who were difficult deliveries present with severe indirect hyperbilirubinemia, an adrenal hemorrhage has to be considered for the aetiology and the cardiovascular status, blood sugar, and electrolytes have to be monitored.

USS-abdomen is the first-line imaging modality in an adrenal hemorrhage; absent color doppler signals over the lesion, liquefaction with mixed echogenicity, or hypoechoic cystic appearance are features of a hemorrhage[6]. However, differentiation of cystic neuroblastoma from an adrenal hemorrhage is challenging. If the USS-abdomen is not conclusive, a CECT-abdomen/chest and urinary VMA levels should be performed to exclude a cystic neuroblastoma[4].

Mutlu et al, in a retrospective study of 13 neonates with adrenal hemorrhage found that it is common among term (62%) and male (77%) neonates and that the hemorrhage resolves in about 4-16 weeks of life. Hence, serial ultrasound scans of the abdomen till the lesion disappears would prevent unnecessary interventions once an adrenal haemorrhage is confirmed[1]. Our case is also a term baby boy and he is followed up with serial ultrasound scans until the complete resolution of the lesion. In conclusion, this case highlights the importance of an ultrasound scan of the abdomen in neonates presenting with severe indirect hyperbilirubinemia to identify an adrenal hemorrhage.

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