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ADRENAL HAEMORRHAGE: CLINICAL PRESENTATION AND ULTRASOUND DIAGNOSIS IN A NIGERIAN NEWBORN

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ABSTRACT

Neonatal Adrenal Haemorrhage (NAH) is a rare condition resulting from a variety of aetiological factors. It has a potential for a catastrophic outcome, thus underscoring the need for a high index of suspicion and a reliable/accurate method of diagnosis. This case report is to alert on the diagnostic possibility of NAH and usefulness of ultrasound diagnosis in the evaluation of a neonate with severe anaemia, non-physiologic hyperbilirubinaemia and abdominal mass.

The subject was a 25 hour old male newborn who was managed for 15 days. He presented with severe anaemia and sepsis for which an exchange blood transfusion was administered. Following a recurrence of the anaemia and palpation of a right hypochondrial mass, he had an abdominal ultra sound evaluation that revealed a 3.47cm by 2.9cm echogenic right supra renal mass. This subsequently became echolucent on serial ultrasonography, presumably following liquefaction. The mass shrunk significantly over the next 15 days with regenerative changes following a period of conservative management. There was no adverse outcome. Final assessment was that of NAH complicated by severe recurrent anaemia and non-physiological hyperbilirubinaemia. The clinical presentation and value of ultrasound diagnosis are discussed.

Keywords: Neonate, Adrenal Haemorrhage, Ultrasound

INTRODUCTION

The adrenal glands can be visualized using ultrasonography in at least 90% of fetuses of more than 26weeks of gestation1. The fetal and neonatal adrenal glands are described as structures having a characteristic V or Y shape[1]. The neonatal adrenal gland is relatively huge, weighing on the average 8g as against the adult weight of 5g[2]. It is also known to decrease in size in infancy as against the increase in size seen in kidneys on serial ultrasound scans[3]. The relatively large size and extreme vascularity of the adrenal glands in neonates make them vulnerable to trauma and traumatic asphyxial injuries, which result in haemorrhagic adrenals[4].

Neonatal adrenal haemorrhage (NAH) is not uncommon in association with birth trauma or infection. The diagnosis may be missed with recovery of the child[2]. The reported incidence of NAH varies from 0.19% to 0.55% on ultrasonography[4-6]. The haemorrhage may present as an abdominal mass, jaundice, anaemia, abdominal calcification[7] or scrotal swelling in male neonates[8]. Symptoms of adrenal insufficiency associated with NAH occur but this is very rare[5]. Where there is a clinical suspicion, the diagnosis is best confirmed by ultrasound. This helps to differentiate NAH from the more solid masses like neuroblastoma[2].

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Despite frequent visualization of the neonatal adrenal glands during ultrasonography, no report of NAH from the tropics was found in the literature reviewed and to the best of the authors' knowledge this is the first reported case of NAH from this part of the world.

CASE REPORT

Baby A.T. presented at 25 hours of age. He was a term, male baby referred to the University of Ilorin Teaching Hospital from a private hospital on account of jaundice and refusal to feed.

Baby was delivered to a 28year old para2+2woman at the referral hospital. Peri-partum history was normal. Mother had no peripartum pyrexia, no premature rupture of membrane and the liquor was not meconium stained. The delivery was spontaneous and vertex. The labour lasted for 3hours. Documented Apgar scores were 7 and 10 at one and five minutes respectively. The weight at birth was 4kg baby was said to have started refusing to suck from 14 hours of age and developed jaundice within the first 24hrs of life.

Physical examination at presentation revealed an ill baby that was markedly pale, having an axillary temperature of 37.2°C with no evidence of respiratory distress. The respiratory rate was 48 cycles per minute. No abnormality was found in the cardiopulmonary system. Also no organ was palpable on initial abdominal examination. Mild head lag,

shoulder girdle hypotonia and weak suckling were observed as deficits on central nervous system examination.

Initial laboratory investigations showed normal haematological indices, except for the PCV of 20% and the fifth Serum electrolytes, urea, creatinine and bilirubin were also within normal limits. However, blood culture yielded Escherichia coli. A single volume exchange blood transfusion (EBT) was carried out with fresh O-positive whole blood to correct the anaemia. There was no ABO incompatibility set up.

At the age of 40hrs (i.e. 15hrs post admission EBT), the PCV remained low at 20%. Examination revealed that baby remained ill, afebrile, with sub-conjunctival haemorrhage. Additional finding on examination of the abdomen was that of a palpable abdominal mass and oligouria. The mass was in the right upper quadrant extending from the right hypochondrium to the right lumbar region. The mass measured 5.0 by 3.0cm in size and it was oval in shape. Abdominal ultrasound showed an echogenic right adrenal mass which measured 3.47cm by 2.9cm in size (Fig. 1). Baby had a straight transfusion which raised the PCV to 40%. Fluid restriction was eased off and the baby started making adequate urine.

The adrenal mass was monitored clinically and sonographically. Serial serum Na+ and K+ remained within normal limits. There was no evidence of adrenal insufficiency. Between 6th and 10th day of life, the baby had persistent pyrexia, refusal of feeds, severe conjugated hyperbilirubinaemia (total bilirubin was 14.47mg%; conjugated fraction was 9.7mg%) and PCV of 24%. The previously demonstrated right adrenal mass (Fig 1) was now completely echolucent (Fig 2). No evidence of obstructive cause of the conjugated hyperbilirubinaemia or venous thrombosis on ultrasound. The WBC was 11.6 x 10^9/l, Neutrophil=15%, Lymphocytes=85%.

At this point, antibiotic was changed from Ceftazidine to Cefuroxime and gentamycin and the baby had another round of EBT done for the correction of the anaemia in order to avoid volume overload. The baby had remarkable improvement after these measures, with all the symptoms subsiding. The patient was discharged home at the age of 17 days. Subsequent ultrasound scanning of the abdomen showed significant reduction in size and areas of regeneration in the right adrenal gland (Fig 3) with PCV stabilizing at 35%. Post discharge follow-up visits on three occasions showed a sustained improvement.

The final diagnostic impression was that of right neonatal adrenal haemorrhage and sepsis complicated by severe anaemia, non-physiological hyperbilirubinaemia and renal impairment.

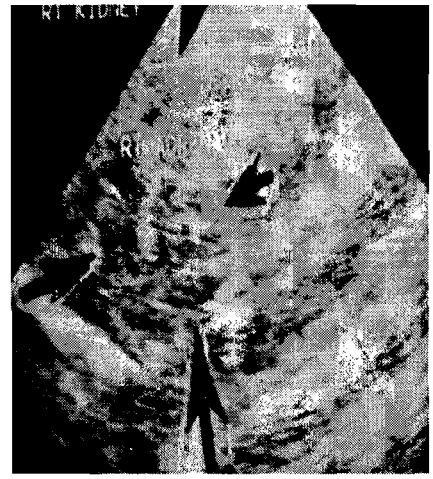

Fig. 1: Ultrasound of the right upper quadrant, showing capsulated echogenic suprarenal mass (arrows). Early stage of right adrenal haemorrhage.

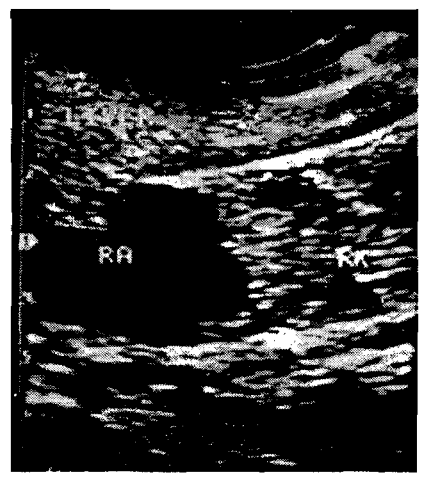

Fig. 2: Ultrasound of the right upper quadrant of the same patient as in Fig. 1, showing complete liquefaction of the right adrenal haemorrhage. It measures 45x36mm in size. RA=Right adrenal; RK=Right Kidney.

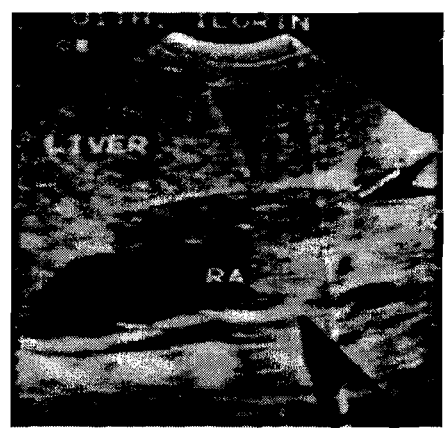

Fig. 3: The same patient in Fig.2, 10 days after fig 2. And there is significant reduction in size of the liquefied (echolucent) central portion, with peripheral area of regenerated solid tissue (arrow heads). The whole gland measures 32x25mm in size. RA=Right adrenal; RK= Right kidney.

DISCUSSION

The very low cases of NAH in the literature to date[4-6], attests to its relative rarity. This coupled with coupled with limited capacity for routine ultrasound examination of the newborn may further contribute to the paucity of information on this condition from developing countries.

Stress and trauma associated with delivery are predisposing factors for adrenal haemorrhage in neonates[2-4]. There was no documented history of asphyxia. The possibility of this and the confirmed sepsis could have been contributory to the development of this condition in this baby. Stressful conditions to the adrenal glands are thought to predispose to greater risk for haemorrhage because of increased stimulation by Adrenocorticotrophic hormone (ACTH). More so, it has been shown in experimental animals that, over stimulation of adrenal glands with ACTH produce degenerative changes and occasionally focal or diffuse haemorrhage in the adrenal cortex[9].

Previous reports indicate that NAH are largely right sided, with male preponderance and occurring in term babies with birth weight between 3-4kg[4,5]. However, the occurrence of adrenocortical insufficiency is rare[4]. Conforming to this reported observation, our case is that of a right sided adrenal haemorrhage in a term, male neonate, weighing 4kg at birth. Throughout the period of follow up the haemorrhage remained intra-capsular, see Fig.1-3. This explains why there was no evidence of scrotal swelling with NAH and was thought to be secondary to haemoperitoneum[8].

Since NAH often causes upper quadrant abdominal mass, the initial clinical impression may be that of renal mass or other non haemorrhagic adrenal tumours. The diagnosis is confirmed by ultrasound which initially like haematoma everywhere show a heterogeneous echogenic mass localised to the adrenal gland (Fig.1) that liquefies with time (Fig.2)[10]. The diagnostic difficulty is with neonatal neuroblastoma. In the case of neuroblastoma presented by Jojart and colleague[7], the tumour behaves sonographycally like haemorrhage. However, it metastasizes to the liver, shows no change in size throughout the 2 months in two neonate with neuroblastoma[11], sonographic picture of mixed echogenicity as distinct from NAH where the haemorrhage may explain why the time. The need for the evolution of the haemorrhage may explain why the adrenal mass was initially not palpable clinically until 15hrs post admission when it became palpable. This would be similar to the evolution of the subperiostal bleed in cephalohaematoma. Other reported additional differentiating feature is by demonstration of blood vessel in neuroblastoma on colour or power doppler examination while NAH appears cystic[12].

The regeneration process noted in our patient (Fig.3) started from the periphery of the gland; this was thought to be as a result of inadequate vascularisation of the innermost adrenocortical cells and the medulla in the repair process. Previous studies in laboratory animals on adrenal regeneration process showed similar changes in the early stage[13].

The management of NAH is conservative[4,5,14,15] as demonstrated in our case, only few need adrenalectomy[6]. The occurrence of anaemia and hyperbilirubinaemia are due to the extra vascular blood in NAH. The option of an EBT therefore provides opportunity for correcting both conditions. Apart from severe anaemia, jaundice and haemoperitoneum, other rare but complications of NAH are cerebral/renal venous thromboses[16,17], adrenocortical insufficiency[5,6] and scrotal swelling[18-20].

In a neonate with unexplained severe anaemia and abdominal mass with sepsis or background perinatal asphyxia, early ultrasound screening followed by serial scan may lead to the diagnosis of adrenal haemorrhage.

Ultrasonography is a cost-effective and simple technique of making a diagnosis of NAH. When diagnosed, NAH usually resolves on conservative management obviating the need for extensive investigations involving ionizing radiation or unnecessary laparatomies.

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