

Unilateral Adrenal Hemorrhage Accompanied by Subgaleal Hematoma with Severe Neonatal Anemia — Case Report

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Abstract

Neonatal adrenal hemorrhage (NAH) occurs in up to 0.2% - 0.55 % of live birth. It is important to note that approximately 10% of these cases might have bilateral involvement. Various risk factors have been reported in addition to birth asphyxia such as sepsis, coagulation disorder, traumatic delivery, and perinatal injuries. However, in a substantial proportion of cases the etiology of bleeding cannot be established. In most cases the event is asymptomatic but in others it may be so devastating and can lead to prolonged adrenal insufficiency and severe cases to shock if not recognized early. This case describes a term infant born with severe anemia in the setting of unilateral adrenal hemorrhage and subgaleal hematoma without resulting in adrenal insufficiency. The infant was successfully treated with blood transfusions and supportive treatment. This is a unique presentation of NAH as it was unilateral associated with right subgaleal hemorrhage and presented with severe anemia.

Keywords: 1-NAH (Neonatal adrenal hemorrhage) 2- AI (Adrenaline insufficiency) 3- IPPV (Intermittent positive pressure ventilation) 4-NICU (Neonatal intensive care unit) 5- USG- (Ultrasonogram) 6-NBL(Neuroblastoma)

Key Learning Points

- Suspect neonatal adrenal hemorrhage in cases of unexplained severe neonatal anemia at birth.
- Adrenal insufficiency is a rare complication of neonatal adrenal hemorrhage.
- It is important to have a conservative approach and avoid early surgery.
- Serial ultrasonography is the preferred modality of investigation.

Introduction

Neonatal adrenal hemorrhage (NAH) is a relatively uncommon condition and occurs in 0.2-0.55 % of infants and is the most common adrenal mass in newborns. It is most common seen in term male infants having higher birth weight. Several factors make adrenal gland prone to hemorrhage which includes relatively large size of adrenal gland to body weight and increase vascularity of the gland which may result in vulnerability to mechanical compression and sensitivity to changes in venous pressure during delivery.

In addition, any factor leading to hypoxia may result in redistribution of blood toward the central nervous system, heart, and adrenal glands. This increase pressure and resultant hypoxia damages the endothelial cells of adrenal gland resulting in adrenal hemorrhage.

Various risk factors have been reported in addition to birth asphyxia, such as sepsis, coagulopathy, birth trauma, and perinatal injuries. A retrospective study identifies vaginal delivery, macrosomia and fetal acidemia as the most important risk factors for NAH. However, in a sizable proportion of cases the etiology of bleeding cannot be established.

Clinical presentation of NAH varies from most commonly asymptomatic to fulminant hemorrhage and shock. The most common presentation is palpable mass in abdomen. Clinical symptoms include jaundice (indirect hyperbilirubinemia resulting from hemorrhage), anemia, lethargy, poor feeding, vomiting, hypotonia and scrotal hemorrhage/scrotal edema. Hypotension and hypoglycemia and hypotonia can present due to adrenal insufficiency.

Almost 80% of NAH are right sided and 5-10% are bilateral. The right adrenal gland is more prone for hemorrhage due to direct drainage of blood flow into the inferior vena cava making it more prone to pressure changes. Also given its anatomy as it lies between the liver and spine which can make more susceptible to compression resulting in increase of venous blood pressure and can lead to hemorrhage.

The neonatal adrenal gland has remarkably high regenerative capacities. NAH usually resolves in 3- 9 months and average resolution time of 18 weeks (about 4 months). Additionally, NAH rarely progresses to Adrenaline insufficiency (AI) since the hemorrhage mostly occur in subscapular region and AI does not occur until 90% of adrenal tissue is destroyed. A minimum of 10% functioning cortisol producing gland is sufficient to prevent progression to AI. Even In the cases of bilateral adrenal hemorrhage, it is less likely that both glands are impacted to the same degree.

The most common imaging modality of choice is serial ultrasound. Computed tomography (CT) and magnetic resonance (MRI) are useful in confirming the presence of hemorrhage and disease progression, but they usually do not provide additional information.

The treatment of NAH conservative management and careful monitoring. Watchful waiting is needed in majority of cases. Interventions are typically not needed unless complications arise. Otherwise, treatment is focused on the clinical outcomes of NAH. Anemia although rare is treated with blood transfusion, while AI is treated with steroids.

Differential diagnoses of lesion near or at the adrenal gland apart from adrenal hemorrhage include adrenal cyst, adrenal abscess, neuroblastoma (NBL) or other solid tumors, congenital adrenal hyperplasia (CAH), pulmonary sequestration, bronchogenic cyst, splenic cyst and cystic lymphangioma.

We are presenting a case of unilateral adrenal hemorrhage with severe anemia without adrenal insufficiency which had a favorable final outcome.

Case Presentation

A male infant was born at term to Primi mother (39 weeks and 5 days of gestation) by vaginal delivery assisted by ventouse. Mothers GBS status was normal and otherwise normal antenatal history (no gestational diabetes or other complications). Antenatal screening was negative. Blood group of mothers was A+ve and liquor was clear. There was no history suggestive of premature rupture of membranes or chorioamnionitis. labor delivery was complicated with prolonged 2nd stage and ventouse was applied to assist the delivery. Baby was born limp with heart rate >60 but no breathing efforts. Baby was resuscitated with suction and IPPV. The baby cried well after successful resuscitation but started shortly with grunting, nasal flaring and tachypnea shortly after birth. The baby was hooked to CPAP and shifted to NICU for post resuscitation care and management. Apgar was 2, 7 and 10 min at 1, 5 and 10 minutes, respectively. Birth weight was 3.820 kg. (Appropriate for gestational Age). Cord blood gases were normal. On examination of baby had a large chignon over the occipital region and other systemic examination of the baby was normal. No abdominal mass or lump was noted on clinical examination. No obvious dysmorphism or congenital anomalies were noted. Baby was hooked to high flow nasal canula with 5 lt of O₂ (FIO₂ 30).

The baby gradually improved and nasal canula was weaned and stopped after 2 hrs of birth as the baby improved. Septic screen (CBC, CRP and blood culture) was taken, and iv fluids (10%Dextrose) and iv antibiotics commenced. On day 2 of birth the baby was looking hypoactive and pale with poor feeding. Further investigation revealed falling Hb - 9.5gm/dl and hematocrit 38.3, crp-5.92mg/dl, normal total and differential count. Metabolic panel was unremarkable and serial blood glucose was normal. Capillary blood gas was normal. Coagulation profile was normal. Blood transfusion was given with packed cells. X-ray chest was unremarkable for cardiopulmonary changes. USG abdomen and USG neurosonogram were done to ascertain the cause of anemia. USG abdomen/color doppler on day 2 was significant revealing a large well defined round anechoic cystic lesion noted in right suprarenal region measuring 2.3 x2.2 cm. Echogenic debris is seen along the periphery of this lesion. No evidence of vascularity seen. The left suprarenal gland was well visualized and appears normal. USG neurosonography and soft tissue skull was suggestive of subgaleal hematoma in the right parietal and right occipital region. The rim collection measures approximately 1.5 mm in thickness. There was no evidence of extension of hematoma across the midline. All labs were repeated including serum cortisol level on day2 which was unremarkable. (serum cortisol 37.8 ug/lt) The hemoglobin improved to 12.9 g/dL and hematocrit improved to 38.5 after his first transfusion. The baby started to improve over the days, reaching full feeding on day 5. Iv antibiotics were stopped on day 4 as blood culture no growth was detected after 72 hrs. Comprehensive Metabolic panel, CBC, and CRP were monitored serially including cortisol level which were within normal limits.

The patient did not require further blood transfusions throughout the remainder of his hospitalization. Repeat USG abdomen was done on day 4 revealed that the right adrenal hemorrhage has slightly increase in size. The lesion was partly anechoic with organizing debris. No evidence of vascularity was seen. The left adrenal gland was normal. On day 7 the USG was repeated, and a follow up scan of right adrenal revealed the hematoma appears increasingly cystic with reduced peripheral echogenic component suggesting resolving hematoma. The patient was discharged against medical advice on day 7 due to social and personal reasons. The baby was discharged on oral Iron drops and vitamin drops.

Blood Analysis of the Newborn

	1 st day of life	Day 2	Day 5	Day 7
CRP (mg/dl)	0.06 mg /dl	5.92 mg/dl	4.5 mg/dl	4.2 mg/dl
WBC COUNT	14.45 k/ul	15.45 k/ul	18.28 k/ul	17.40 k/ul
Hb (g/dl)	10.5 gms/dl	9.5gms/dl	12.9 gms/dl	12.5 gms/dl
HCT	34.60	28.3	38.5	37.5
Na mEq/L	135.1mmol/lt (136-145 mmol/lt)	138.7 mmol/lt		137 mmol/lt
K mEq/L	3.84 mmol/lt	4.64 mmol/lt		4.5 mmol.lt
PT		12 sec		
APTT		30 sec		
AST U/L	17.21	20.40		19.45
ALT U/L	38	40		
Serum total cortisol		37.8 ugm/lt (23.2 -194.3ug/lt)		39.8 ugm/lt

Ultrasound Images

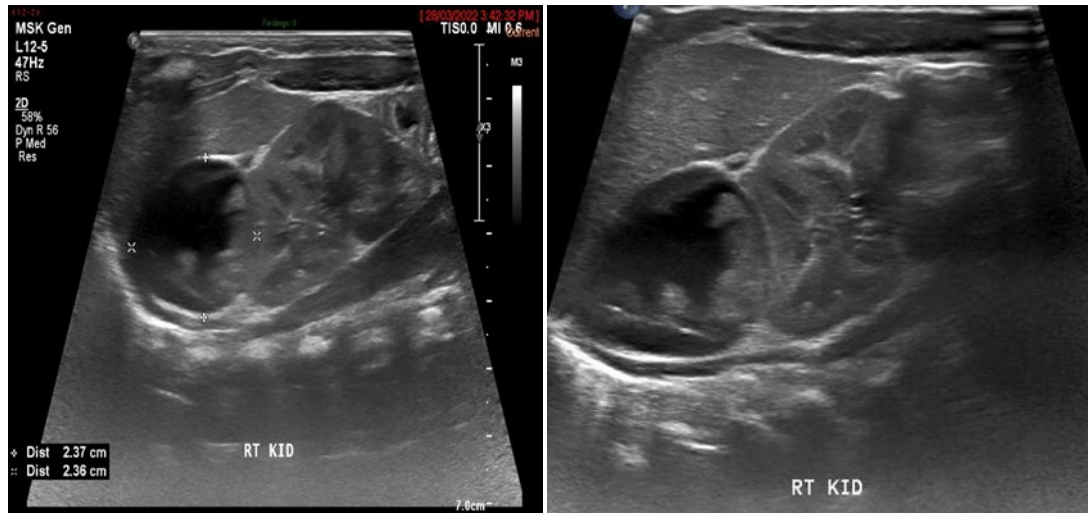


Figure 1&2 : Cystic lesion in the right suprarenal region with an anechoic fluid component and partial echogenic peripheral component in keeping with hematoma. The right kidney is seen separate from the lesion



Figure 3: Follow up scan of right adrenal hematoma after 4 days. The hematoma appears increasingly cystic with reduced peripheral echogenic component suggesting resolving hematoma.



Figure 4: Thin rim of fluid collection with thin internal septations seen in the right parietal region superficial to the underlying bone, not crossing the midline suggesting subgaleal hematoma.

Discussion and Conclusion

While literature has provided variable clinical presentation of adrenal hemorrhage there are multiple case reports demonstrating anemia as a common clinical presentation. Like other case studies that demonstrate that male gender preference for NAH, our patient was male and displayed anemia, hypotonia, and poor feeding on physical examination. Our patient also had risk factors associated with NAH such as a traumatic or prolonged delivery, an assisted birth with ventouse and perinatal asphyxia requiring resuscitation at birth.

Adrenal hemorrhage should be on the differential when an infant is born with clinical and laboratory signs of anemia with no obvious cause. Although rare, any neonate with adrenal hemorrhage should be closely monitored for adrenal insufficiency.

Serial USG is the most reliable radiological modality for monitoring of adrenal gland hemorrhage. When repeated over time, it may allow for monitoring of the evolution of changes and for differentiation with other causes such as malignant and benign tumors. When differentiated from NBL, hemorrhage showed no vascular flow in color Doppler US and gradual regression of lesions over time.

With appropriate clinical investigations, watchful waiting, Supportive treatment and treatment of anemia with blood transfusion is all needed in majority of the cases. In rare cases with a baby having adrenal insufficiency should be treated with iv hydrocortisone and pediatric endocrinology opinion and referral should be taken.

Declaration of Interest

The authors declare that there are no potential conflicts (financial, professional, or personal) relevant to the content presented in this article.

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