Case Report

Neonatal adrenal hemorrhage presenting as an acute scrotum: A case report on the rare presentation of right adrenal hemorrhage and contralateral left scrotal hematoma

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Abbreviations & Acronyms
AH = adrenal hemorrhage
CT = computed tomography
MRI = magnetic resonance imaging
NAH = neonatal adrenal hemorrhage
SH = scrotal hematoma
US = ultrasonography

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Introduction: Acute swelling and discoloration of the scrotum in a newborn is a rare condition and can have several causes such as testicular torsion, trauma, inguinal hernia, hydrocele, or adrenal hemorrhage.

Case presentation: We report a neonate of adrenal hemorrhage presenting clinically as the acute scrotum. Definitive diagnosis was defined by ultrasonography and computed tomography scan, and the conservative management was successfully performed.

Conclusion: Adrenal hemorrhage should be considered as one of the causes of acute scrotum in newborns. The abdominal ultrasonography, as well as the scrotal ultrasonography, should be performed routinely to achieve a definitive diagnosis to avoid unnecessary invasive procedures.

Key words: acute scrotum, adrenal hemorrhage, conservative treatment, newborn, ultrasonography.

Keynote message

The acute scrotum during the neonatal period represents an emergency due to the viability of the testis. Neonatal adrenal hemorrhage (NAH), although rare, should be considered in scrotal swelling with discoloration in newborns. NAH was generally treated conservatively and has a favorable prognosis. Abdominal ultrasonography (US), as well as scrotal US, should be performed routinely in case of acute scrotum in newborns for definitive diagnosis to avoid unnecessary invasive procedures.

Introduction

Acute scrotum is a clinical condition represented by sudden pain and/or swelling. In newborns, the occurrence of an acute scrotum is a surgical emergency because the viability of the testis is involved.[1] SH in a newborn is a rare condition but warrants prompt diagnosis and urgent intervention. It commonly results from testicular torsion, birth-related trauma, and rarely AH. However, in some cases, the cause was idiopathic, and scrotal exploration was considered necessary for the diagnosis and treatment of these neonates.[2]

NAH is a rare condition with an incidence of 0.2%,[3] and SH is an extremely rare manifestation of NAH.[4]

Herein, we report a rare case of NAH with right AH and contralateral left SH that was managed conservatively.

Case presentation

A 3450-g male was born by normal vaginal delivery in a cephalic position after 39 weeks of gestation with an uneventful pregnancy. No history of birth trauma was present. Apgar score was 9 at the first minute and 9 at the fifth minute. On his first day of life, a swelling and bluish discoloration of the left scrotum were noted (Fig. 1), and the infant was transferred to our hospital. On physical examination, there was no abdominal distention and the scrotal swelling

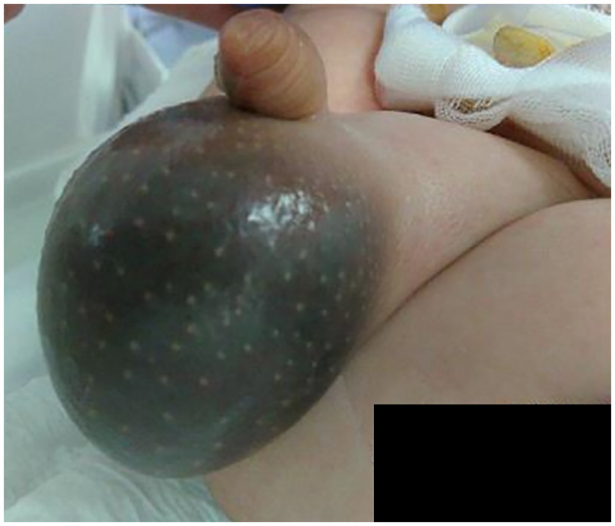

Fig. 1 Left scrotal swelling with bluish discoloration.

was tender. At first, we had a suspicion of testicular torsion with an impression of something atypical. The scrotal US including power Doppler showed that the left testis maintained normal echogenic structure and vascularity, and the hematic fluid correction in the left side of the scrotum (Fig. 2a). The abdominal US revealed hematoma in the inferior liver region (Fig. 2b). A subsequent abdominal CT scan revealed a right adrenal hematoma, but no obvious hemoperitoneum (Fig. 3). The hemogram results were hemoglobin 9.8 g/dL, and platelets 234.000/mm^3. Blood coagulation tests were normal. Biochemistry revealed total bilirubin 5.4 mg/dL and direct bilirubin 0.5 mg/dL. Fresh frozen plasma (2 IU/day) and vitamin K (2 mg/day) were administered on the second and third days of life. The hemoglobin level increased to 14 g/dL on the seventh day of life. The infant was discharged home on the 14th day of life in good condition. The swelling of the scrotum had disappeared on the day of discharge. The infant visited the outpatient clinic every 2 months for 6 months after discharge, and MRI at 6 months revealed both adrenal and scrotal hemorrhage had disappeared.

Discussion

NAH is associated with perinatal hypoxia, difficult delivery, shock, septicemia, and coagulation disorders.[5] The adrenal gland in newborns is very large and vulnerable to vascular damage.[3] The right adrenal gland is the frequent site of NAH.[5] It easily becomes trapped between the liver and spine, causing hemorrhage. In addition, the right adrenal vein drains generally directly into the inferior vena cava and is exposed to changes in venous pressure.[6] Most NAH was treated conservatively and had a favorable prognosis with complete resolution within 6 months without any complications,[7] whereas a fatal case due to hemorrhagic shock has also been reported.[8]

The etiology of SH could consider that the hemorrhage from the adrenal gland spreads to the retroperitoneal or peritoneal cavity, and the blood reaches the scrotum through a

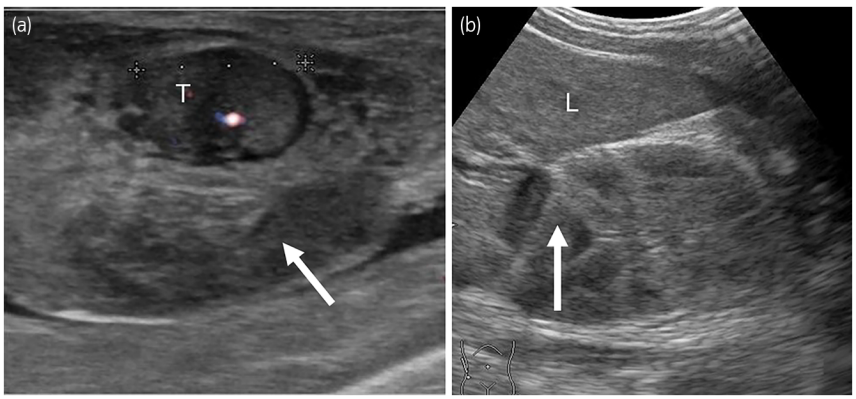

Fig. 2 (a) Scrotal US of the left side of the scrotum showed hematic fluid correction in the left scrotum (white arrow). (b) Abdominal US revealed hematoma (white arrow) in the inferior liver region (T: testis, L: liver).

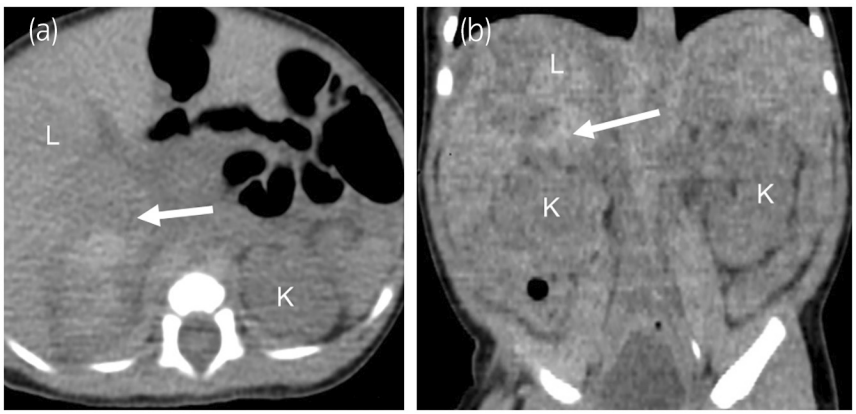

Fig. 3 CT scan (a: axial section, b: coronal section) revealed right adrenal hematoma (white arrow). (K: kidney, L: liver).

Table 1 Laterality and treatment of neonatal AH associated with SH of 13 cases reported since 2012 (including our case)

Side of AH	Cases	Side of SH	Cases
Right	10	Right	8
		Left	2
Left	1	Right	1
Bilateral	1	Right	1
Unknown	1	Bilateral	1
Treatment	Cases		
Conservative	11		
Surgical	2		

patent processus vaginalis or by dissection of the tissue of the retroperitoneum.[3]

Lai *et al*. reviewed 29 cases of NAH with SH reported until 2011,[4] since then 12 cases in the literature have been reported.[4,8–17] Of the 12 cases of NAH with SH, 8 out of 12 cases (66.7%) were presented as right AH with right SH, and surgical treatment was performed in two cases (16.7%) (Table 1). Although there is a growing number of reports of NAH with SH, the onset of SH on the opposite side of AH is less frequent. Our case is the rare presentation of NAH with right AH and contralateral left SH as is the case reported by Maximiano C *et al*. The possible mechanism might be as follows: the right AH spreads to the peritoneal cavity and the presence of a left patent processus vaginalis allowed the passage of blood into the scrotum.[9]

Acute scrotal discoloration in neonates is rare but usually caused by testicular torsion which might be a surgical emergency. Although surgical exploration was considered the gold standard, unnecessary surgical exploration has often been performed because of diagnostic uncertainty.[4]

Apart from NAH, splenic hemorrhage and subcapsular liver hematoma have been reported as causes of SH, which are rare conditions in neonates.[18,19] They may be life threatening due to hemorrhagic shock, especially in case of splenic rupture or rupture of the subcapsular liver hematoma. Therefore, it should be taken into account that SH can act as a window for intra-abdominal hemorrhage.

The US is a useful modality for detecting AH as well as testicular torsion as it can be carried out in an emergency setting without invasive procedures and radiation exposure.

In fact, US is the criterion standard modality for both the initial screening and the follow-up evaluation in neonates, and is very specific in differentiating the major causes of adrenal masses. Other imaging techniques such as CT or MRI are useful in confirming the presence of hemorrhage and progression of hemoglobin breakdown.

Conclusion

In a newborn with an acute scrotum, NAH should be considered in the differential diagnosis. As a routine examination, the abdominal, as well as scrotal, US should be performed to achieve the definitive diagnosis in neonates with scrotal swelling and bluish discoloration for avoiding unnecessary invasive procedures.

Conflict of interest

The authors declare no conflict of interest.

Approval of the research protocol by an Institutional Reviewer Board

Not applicable.

Informed consent

We obtained consent from the family for the presentation of this case.

Registry and the Registration No. of the study/trial

Not applicable.

Author contributions

Takashi Okamoto: Data curation; writing – original draft. Shinya Kajiwara: Data curation. Sho Sekito: Data curation. Takuji Shibahara: Data curation. Takehisa Onishi: Conceptualization; writing – original draft; writing – review and editing.

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Editorial Comment

Editorial comment from Dr. Alonso, Dr. Perez-Bertolez, and Dr. Castro to neonatal adrenal hemorrhage presenting as an acute scrotum: A case report on the rare presentation of right adrenal hemorrhage and contralateral left scrotal hematoma

Neonatal adrenal hemorrhage (NAH) is an infrequent condition (1.7–2.1/1000 births).[1] It is more common in boys and on the right side (70%) as in the case reported by Okamoto et al.,[2] where it can be compressed between the liver and spine. It is remarkable that adrenal glands in newborns are approximately 10–20 times bigger in comparison with adults relative to body weight, increasing their vulnerability.[1]

As it is explained by these authors,[2] NAH is associated with difficult delivery, shock, septicemia, coagulopathy, and any factor leading to hypoxia and redistribution of blood toward the central nervous system, heart, and adrenal gland. Spontaneous and prenatal occurrences are also documented.

Manifestations depend on the degree of hemorrhage and adrenal cortex compromised. The most frequent presentations are anemia, jaundice, abdominal distention, and flank mass.[1] The scrotal discoloration (Bryant's sign), seen in this patient, and inguinal ecchymosis (Stabler's sign) are unusual. Nevertheless, when they are present, we should suspect retroperitoneal hemorrhage.

Hemogram, biochemistry, and coagulation tests are required. Clinical and ultrasonography (US) follow-up is mandatory for the assessment of hemorrhage resolution and conservative management.

We think that the magnetic resonance imaging performed to this patient was not necessary after the diagnosis of NAH because it does not provide additional information. However, it may help at first to differentiate this etiology from neonatal cystic neuroblastoma.

What is noticeable about this case is the presence of NAH with contralateral scrotal hematoma, which is poorly reported. Additionally, the only risk factors were vaginal delivery, male gender, and being a term neonate.

On the other hand, scrotal hematoma often raises the suspicion of testicular torsion. Perinatal testicular torsion is unusual (6.1/100,000 births). Most of them (75%) undergoing prenatally. Physical examination reveals firm, erythematous swollen testicles that do not transmit light. US shows an enlarged testis and epididymis.[3]

To conclude, we agree with the authors that NAH should be ruled out in cases of acute scrotum, mostly after a difficult delivery and/or asphyxia. A US is the first step, since it does not necessitate neither radiation nor sedation. An integration of clinical information, physical examination, and US is necessary to achieve the diagnosis. This association allows conservative treatment, avoiding a surgical exploration.

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Conflict of interest

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