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Adrenal abscess in a preterm neonate with sepsis

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ABSTRACT

The adrenal abscess is a rare complication of adrenal hemorrhage in the neonatal period. Due to its rare occurrence and non-specific signs, diagnosing and treating an adrenal abscess in the neonatal period might be challenging. We present herein a 3-week-old male neonate with an adrenal abscess associated with *Escherichia coli* sepsis, which was successfully treated by open surgery (using the minimal posterior lumbar approach) following an unsuccessful ultrasound-guided percutaneous drainage.

1. Introduction

A neonatal adrenal gland abscess is a rare pathology, with some 50 cases reported in the literature so far [1]. Consequently, it is rarely included in the differential diagnosis of an adrenal mass. Adrenal abscesses usually develop within the adrenal hemorrhage, associated with a traumatic or difficult delivery, hypoxia, sepsis, and coagulopathy. In addition, adrenal abscesses may develop due to a hematogenic spread of bacteria to “normal” adrenal glands, which is observed in neonates with sepsis [2]. Timely and adequate diagnosis is crucial for the appropriate management and the patient's outcome [2]. Although ultrasound-guided percutaneous drainage is the effective therapeutic approach in most cases, definitive surgery drainage is sometimes indicated [3].

We present a 3-week-old preterm male neonate with an adrenal abscess as a complication of *Escherichia coli* sepsis. The abscess was successfully treated by surgery drainage combined with the antibiotics following repeated ultrasound-guided percutaneous drainage.

2. Case report

A male neonate was referred to our hospital from the county hospital 16 hours following a complicated vaginal delivery caused by prolonged labor. The baby was born in the 35th week with a weight of 3250 g and a length of 48 cm. His Apgar score was six at the first minute and seven at the fifth minute. Physical examination revealed no abnormalities except tachypnea (>60 breaths/minute). Neonate required oxygen supplementation and nasal continuous positive airway pressure in the neonatal intensive care unit (NICU) for two days and had respiratory distress. Reflexes were normal. The blood culture was positive for *Escherichia Coli* on the second

Abbreviations: CRP, C-reactive protein; HVA, Homovanillic acid; MRI, Magnetic Resonance Imaging; NICU, Neonatal intensive care unit; NSE, Neuron-specific enolase; VMA, Vanillylmandelic acid.

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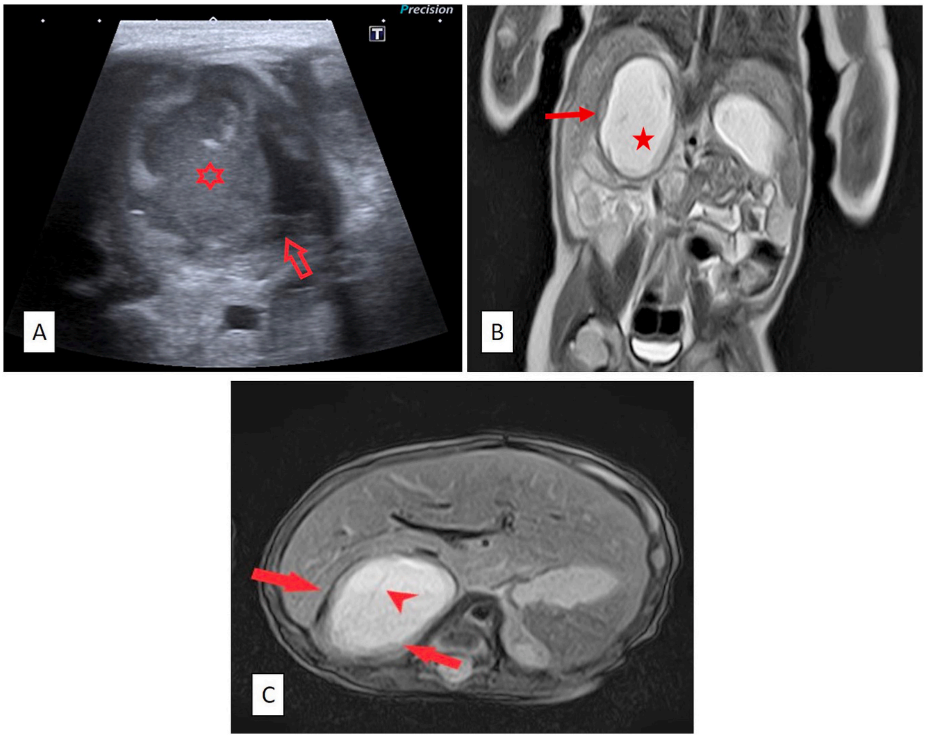

Fig. 1. A-C. (A): Ultrasound image showing a hypoechoic, round lesion filled with fluid (red star) and hypertrophic wall (red arrow); (B): Coronal T2 weighted image showing mostly hyperintense (cystic) lesion in right suprarenal space (red star) with thick, hypointense capsule (red arrow); (C): Axial T2 weighted image showing cystic right suprarenal lesion (red arrows) with thin intraluminal septa (redpointed arrow). (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

postpartum day. Cerebral fluid cultures were negative. The treatment with Meropenem was initiated immediately. Despite stabilization of the respiratory distress, the baby remained febrile (38.4 °C) and irritable. In addition, leukocytosis persisted (18.5 thousand/mm^3), whiles the C-reactive protein (CRP) increased from 45.6 mg/dl to 146.2 mg/dl, prompting a search for occult infection. On the seventh postpartum day, an abdominal ultrasound showed a right, hypoechoic, suprarenal lesion filled with thick fluid and hypertrophic wall. The lesion was predominantly avascular and cystic, measuring 41 × 37 × 48 mm (Fig. 1A). Magnetic Resonance Imaging (MRI) of the abdomen revealed a hyperintense (cystic) lesion in the right suprarenal space with thick, hypointense capsule and intraluminal septa (Fig. 1B–C). Clinical and radiological findings were suggestive of an adrenal hemorrhage complicated by secondary infection and abscess formation. All other laboratory tests [Coagulation profile, renal and liver function tests, neuron-specific enolase (NSE), Vanillylmandelic acid (VMA), homovanillic acid (HVA), and cortisol] were all within the normal range. Two unsuccessful treatments with ultrasound-guided percutaneous drainage were performed. The drainage was not possible due to septation of the abscess, thickening of the capsule, and inability to completely evacuate the purulent fluid. Consequently, the adrenal mass was drained by open surgery on the 21st postpartum day. The surgery was performed using the minimal posterior lumbar approach. Fifty ml of purulent fluid was removed. The aspirate cultures were positive for Meropenem-sensitive *E. coli*. The tube drain was removed on the sixth postoperative day. The recovery went without complications. The elevated infection laboratory parameters normalized (leukocytes and CRP), and the patient was discharged after 14 days of the treatment. At six months follow-up, the child was doing well with no clinical signs or symptoms of adrenal insufficiency.

3. Discussion

This case highlights the importance of the adequate management of the neonatal adrenal abscess. Adrenal abscess in the neonatal period is challenging to diagnose as the condition is exceedingly rare and can present with non-specific signs and symptoms. Because of the specific structure of this organ, it is considered to develop mainly because of superinfection and inflammation within the site of previously extravasated blood to the adrenal gland [3,4]. It should also be noted that adrenal glands in neonates are relatively large (persistent fetal cortex) and well-vascularized.

The broad availability of ultrasonography in neonatal units enables adrenal hematomas to be detected early. It is estimated that they develop in 1.7–2.1 per 1.000 neonates [4]. They are statistically more frequent in term-born neonates with significant perinatal history (e.g., birth trauma, perinatal hypoxia, intrauterine infections, and coagulation disorders) [2,4]. Due to anatomic differences, hemorrhage is more frequent (70%) in the right adrenal gland, a phenomenon thought to result from compression of the gland located between the liver and the right kidney [4,5].

The differential diagnosis of cystic adrenal masses is wide, including hemorrhage, cysts, neoplasms (neuroblastoma, nephroblastoma/Wilms' tumor/), renal duplication with dilatation of the upper segment, and hydronephrosis [6,7]. In our case, the clinical presentation and course were strongly suggestive of an infectious condition.

Two approaches have been proposed regarding the development of a neonatal adrenal abscess: (1) Hematogenous bacterial seeding of a normal adrenal gland and (2) seeding of an adrenal hemorrhage with subsequent abscess formation [2]. In most cases, adrenal abscesses result from adrenal hemorrhage associated with a traumatic or difficult delivery, hypoxia, sepsis, or coagulopathy [2,8–10]. We believe that our case was due to adrenal hemorrhage with secondary bacterial seeding caused by sepsis (Blood culture was positive for *E. coli*). Notably, adrenal hemorrhage may be bilateral in ~10% of cases [11].

Neonatal sepsis is a potentially lethal condition, particularly in preterm neonates, in which *E. coli* is a leading causative microorganism. Early-onset neonatal sepsis is associated with the acquisition of microorganisms from the mother. In our case, we had no clear evidence of the maternal source of the infection. This may be caused by the neonate being referred to our hospital from another (county) hospital. Risk factors for *E. coli* infection include low gestational age, intrapartum fever, and prolonged rupture of membranes [12].

In conclusion, a rapid, early, and accurate diagnosis of the adrenal abscess is crucial for appropriate treatment to prevent a generalized infection and destruction of adjacent organs. Although neonatal adrenal abscess is uncommon, it should be considered in a differential diagnosis of septicemic neonates with an adrenal mass, particularly those with a history of perinatal hypoxia with prolonged labor.

4. Patient consent

Consent to publish the case report was obtained from the patient's mother.

Authorship

All authors attest that they meet the current ICMJE criteria for authorship.

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Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have influenced the work reported in this paper.

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