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Acute Cerebral Venous Sinus Thrombosis as a Complication of Acute Hyponatremic Dehydration in Neonate: Case Report and Mini Review

Anwar Ramadan Alhamad[1*], Yassin Alsaleh[2] and Nabil Almajhad[3]

[1]*Senior registrar of General Pediatrics, Maternal and children hospital in Alhasa, Saudi Arabia*

[2]*Pediatric Endocrinology consultant, Maternal and children hospital in Alhasa, Saudi Arabia*

[3]*Pediatric Neurology consultant, Maternal and children hospital in Alhasa, Saudi Arabia*

***Address for Correspondence:**
Anwar Ramadan Alhamad, Senior registrar of General Pediatrics, Maternal and children hospital in Alhasa, Soudi Arabia;
E-mail: anwr-r-hmd@hotmail.com

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Abstract

Cerebral Sinus Venous Thrombosis (CSVT) is a rare disease with severe neurological sequela. CVST results from genetic and acquired prothrombotic disorders. The diagnosis is difficult and is missed in some cases because of the highly variable clinical presentation in neonate.

Our case report showed a 26-day-old neonate who developed CVST secondary to acute gastroenteritis with hyponatremic dehydration. CVST was confirmed by imaging studies, and the patient was managed symptomatically after excluding other causes.

Introduction

Neonatal Cerebral Venous Sinus Thrombosis (CVST) is a rare disease [1,2] that results in neurologic impairment or death in approximately half of cases [3]. There is a higher risk among boys both for arterial ischemic stroke and for sinovenous thrombosis. The reason for the sex predilection is not clear [1,4]. Its incidence is approximately0.67 cases per100,000 children per year. Neonates are the most commonly affected [5], though the true incidence in neonates is still unknown, because reliable epidemiologic data are lacking [3].

The reported incidence of neonatal SVT is probably an underestimation; many casesmay remain unidentified due to nonspecific clinical presentation. Neonatal sinovenous thrombosis is a multifactorial disease [2,4,6,7] that results from a combination of prothrombotic risk factors and/or underlying clinical conditions [6]. Infections are the most common predisposing factors both in neonates and older children, followed by hypercoagulable/ hematological states, dehydration, and various other conditions [1,8]. Acute systemic illness is the dominant risk factor among newbornsin addition to infection and fluid/electrolyte disturbances [9].

The most common locations for CVST in neonates are the transverse sinuses, the superior sagittal sinus, followed by the straight sinus in the frontal and parietal lobes [7].

Presentations of CSVT are highly variable. Generalized or focal seizures are the most common presentation. However, lethargy, feeding difficulties, apnea or respiratory distress, and hypotonia predominate in neonates [1]. Hemiplegia may occasionally be the presenting sign seen in neonates with CSVT upon examination [10]. New borns may show signs of intracranial hypertension, such as papilledema and sixth-nerve palsy or a bulging fontanelle.

Anticoagulation is the most common specific treatment used. To date, there is no consensus on the role of anticoagulant therapy in children. The treatment dilemma is especially pronounced for neonatal CSVT.

In our case, we presented CVST developed in neonates as a complication of acute hyponatremic gastroenteritis, with severe dehydration as uncommon sequel in order to discuss the difficulties in determining cause and effect as well as emphasize the importance of early evaluation of dehydration to manage it timely and appropriately.

Case Presentation

The patient was a 26-day-old Saudi male neonate, a product of full-term, normal spontaneous vaginal delivery with noperinatal complications. The mother faced no hypoxia at birth, premature rupture of membranes, maternal infection, placental abruption, or gestational diabetes. The patient exhibited no family history of bleeding, thrombosis, or seizure disorder.

He presented with a history of diarrhea and vomiting for 10 days. He was admitted to the ward as a case of acute hyponatremia gastroenteritis with severe dehydration and was managed by Normal Saline bolus and sodium bicarbonate. Biochemistry showed that Serum sodium =130 mmol/L, potassium=4.54 mmol/L, chloride=104.7 mmol/L, bicarbonate 14 mmol/L, blood urea nitrogen=3.9 mmol/L, creatinine=29 µmol/L, glucose =96.8 mg/ dl. After 3 days of admission, the patient suddenly became pale, lethargic, and hypoactive, with mottled skin, delayed capillary refill time (4 seconds), and a bulging anterior fontanel. Also he developed clonic convulsion on the left side of the body, which was aborted after loading dose of phenytoin, Complete Blood Count done urgently, which revealed a drop of hemoglobin from 12.3 to 7g/dl. Coagulation profile and platelet were within normal limits (platelet =804*103/ ul, prothrombin time =12.90sec, and partial thromboplastin time

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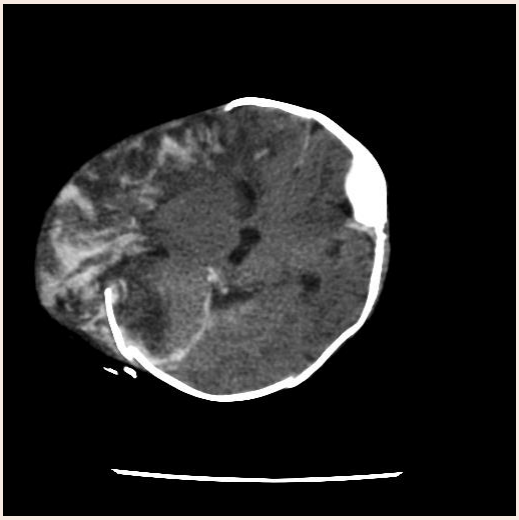

Figure 1: Brain Computer Tomography.

=38.10 sec). Cranial US were done, which showed Intra-Ventricular Hemorrhage grade 1. Due to dissatisfaction with US findings, Brain Computer Tomography was requested.

Brain CT was performed, and the result was right subarachnoid and subdural hemorrhage, infarction, and midline shift with acute venous sinus thrombosis. Contrast revealed a filling defect, which indicates acute venous sinus thrombosis complicated by venous infarction (Figure 1).

The patient was shifted to the Pediatric Intensive Care Unit for more close observation and monitoring and to apply neuroprotective measures. Septic workup was done, and the patient started on antibiotics with supportive care including transfusion of Packed Red Blood Cells (PRBC), platelets, and fresh frozen plasma. No intubation or inotropes were needed.

Magnetic Resonance Arteriogram (MRA) and Magnetic Resonance Venogram (MRV) were done and showed increase in subdural hematoma along the falx and right temporoparietal regions (Figures 2 and 3). An increase in the size of the supratentorial ventrical related to moderate supratentorial hydrocephalus with intraventricular heomorrhage was seen within the dependent portion of the occipital horn of lateral ventricales. Filling defect was still noted with luminal enhancement within the right transverse sinus and superior sagittal sinus related to venous thrombosis.

Craniotomy and evacuation was done by the neurosurgeon

Patient was seen by pediatric hematologist. Protein C,S and free S, Hemocysteine level, Fibrinogen level, Thrombin time, Factor VIII and V, and Antithrombin III were requested, and all came within normal limits except PFA100, which was not available.

Echocardiography was done which showed only mild mitral regurgitation and patent foramen ovals. Convulsions were controlled with phenobarbitone. Later, a ventricular-peritoneal shunt was inserted.

Discussion

Cerebral Venous Thrombosis (CVST) in neonates is a rare but serious disease that is being increasingly recognized, mainly because of the increasing clinical awareness of the disease and availability of more sensitive diagnostic tools. Neonates are more vulnerable to brain lesions, and the lesions tend to be more hemorrhagic in neonates and infants with CSVT [11]. The occurrence of venous infarcts portends a poor outcome [3]. Abnormality in levels of prothrombotic factors is common in neonates. There is controversy regarding whether some of these may be epiphenomena and coincidental vs. causal in nature [9]. The pathophysiologic mechanism of parenchymal injuries related to CSVT is not fully understood [11]. In neonates in addition to the causative factors promoting thrombosis as in older children, the normal moulding and overlapping of the cranial sutures during birth can damage cerebral sinus structures that immediately underlie the sagittal and lateral sinuses promoting CSVT [10].

There was a high incidence of hemorrhage in neonates76%. The reason for this tendency to hemorrhage in patients with CSVT is the raised venous pressure that gets transmitted in a retrograde fashion proximal to the obstruction. This increases the venular pressure and the capillary hydrostatic pressure, resulting in leakage of the capillary fluid into the interstitial space, causing edema. The fluid leakage is almost always accompanied by the “diapedesis” of red blood cells, which is usually the cause of a high incidence of hemorrhagic venous infarcts in CSVT [11].

Morbidity and mortality can, however, be significant and depend on the extent and localization of thrombosis and associated cerebral parenchymal lesions [4,12,13].

Adverse neurological sequela consist of general developmental delay, sensorimotor deficits, visual impairments, and epilepsy [4].

CVST in neonates is unpredictable. Neonates in general have a greater risk of poor outcomes [13].

The presence of infarcts, younger age, seizures at presentation, decreased level of consciousness, focal neurological signs, and thrombosis of the straight sinus have been found to be associated with poor outcome and long-term major neurological deficits in both neonates and older children [1,3,13].

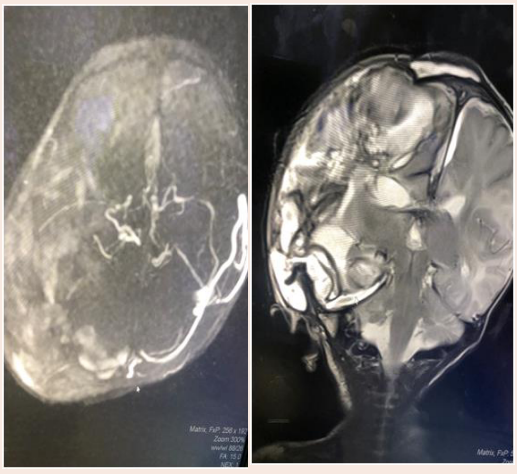

Figure 2 and 3: Magnetic Resonance Venogram.

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Hypernatremic dehydration is a known risk factor for cerebral sinus thrombosis. Hypernatremic dehydration causes a cerebral sinovenous thrombosis, which then leads to a consumptive coagulopathy and secondary widespread intracranial hemorrhage. The sodium level in our patient was low as opposed to what was reported previously.

Reported a case of CSVT resulting from severe hypernatremic dehydration and diabetes insipidus in a breastfed neonate in transverse venous sinus and secondary widespread intracranial hemorrhage after 2 days of hospitalization [14]. Also documented a case of CVST and large thrombus in the abdominal aorta associated with hypernatremia [15].

The clinical presentation varied and was independent of the cerebral sinus affected [1-20]. Presenting symptoms in neonates most frequently include seizures. In one study, convulsion developed after a median time interval of 1.5 days postnatally [4]; in our patient, due to the absence of perinatal risk factors, it presented relatively late, and dehydration was only 1.9% as a risk factor.

The widespread use of neuroimaging now allows for early diagnosis and completely modified our knowledge on this disorder [16].

Cranial ultrasound may be helpful in neonatal cerebral venous thrombosis, but its findings often need to be confirmed by MRI and MRV [1]; in our case, it showed only IVH 1.Also, in neonates, computerized axial tomography is a really not ideal, as it may miss the diagnosis in at least 10% of cases [10]. Afalse positive result may be seen in the non-contrast CT because of a high hematocrit [3]. MRV is considered the technique of choice for diagnosis and follow-up of CVST, but in certain cases, MRI could be superior, as it shows the thrombus itself and not just the absence of signal, as seen on MRV [1]. Berfelo et al. found that Multi sinus thrombosis was present in 9 of 10 newborns who died. Our patient was found to have multi sinus thrombosis.

The management of neonatal thrombosis has no special protocols [15]. The evidence supporting most recommendations for antithrombotic therapy in neonates and children remains weak [11].

Therapy is largely supportive. However, The American College of Chest Physician guidelines, published in 2004 and updated in 2008, suggests anticoagulation for neonates without significant intracranial hemorrhage, while the American Heart Association (AHA) guidelines published in 2008 recommend anticoagulants only when there is evidence of thrombus propagation, multiple cerebral or systemic emboli, or a severe prothrombotic state is present [1]. Our patient was managed without anticoagulant.

Supportive treatment is more or less uniform for all pediatric age groups and includes rehydration, antibiotics for suspected sepsis, antiepileptic drugs for seizure control, and measures to reduce intracranial hypertension [1].

Surgical treatment of cerebral venous thrombosis is generally reserved for intracerebral hematomas with mass effect and for hydrocephalus [1], as in our patient, for whom the craniotomy was done followed by Ventricular-Peritoneal shunt insertion.

CVST in our presented case is attributed to acute hyponatremic gastroenteritis with severe dehydration. So it is vital to consider CVST in differential diagnosis in any neonate presented with seizure and dehydration.

Conclusion

CVST is a serious complication of acute gastroenteritis with hyponatremic severe dehydration in neonates. The course duration of this complication is variable. We present this case to emphasis the importance of close follow up during the first days of admission of gastroenteritis patients, especially those with moderate to severe dehydration.

Given the increasing incidence of sinovenous thrombosis in children, the variations in treatment, and the devastating outcomes in half the children with this disorder, studies are needed to identify more effective immediate and secondary preventive therapies and to be alert to any change in neurological examination to obtain any extension of thrombus.

In Saudi Arabia, the overall incidence of CSVT is unknown. In our case, the most important risk for development of thrombosis is hyponatremic dehydration, which was responsible for the thrombosis.

One needs to be aware of this possibility in neonates and look for it in any suspected term neonate with seizures and dehydration.

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