Coexisting Cerebral Salt Wasting Syndrome and Central Diabetes Insipidus in a Patient with Posterior Cerebrovascular Infarction: A Case Report

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Keywords

Cerebral salt wasting syndrome · Central diabetes insipidus · Hyponatremia · Hypernatremia · Cerebrovascular infarction

Abstract

Background: Disorders of sodium balance are common in critically ill neurologic patients. However, the coexisting of cerebral salt wasting syndrome (CSW) and central diabetes insipidus (CDI) in such patients is rare. Early recognition of such conditions is challenging, thus making the prognosis ominous. **Case Presentation:** A 50-year-old male patient presented with acute posterior cerebrovascular infarction complicated by several attacks of disturbed sodium homeostasis. The first attack manifested as hypernatremia (up to 161 mmol/L) and polyuria with high urine sodium (188 mmol/L) could only be explained by CSW on top of CDI. Especially the patient was not receiving any hyperosmolar or sodium-containing fluids. Serum sodium was corrected by desmopressin acetate. Later, the patient developed 2 attacks of hyponatremia (down to 119 mmol/L) diagnosed as CSW that was treated with fludrocortisone. Finally, he developed

hypernatremia (up to 165 mmol/L) diagnosed as CDI and was treated with desmopressin acetate. **Conclusion:** Sodium hemostasis disorders require full consideration of serum electrolytes, intravascular volume state, and urine electrolytes in view of the clinical condition. Early diagnosis and administration of the proper treatment are the cornerstones of successful management.

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Introduction

Sodium dysregulation is considered one of the most urgent challenging situations in intensive care medicine. Several neurologic disorders including tumors, neurosurgeries, subarachnoid hemorrhages, and traumatic brain injuries could result in sodium abnormality [1]. The sodium dysregulation could be presented as central diabetes insipidus (CDI), cerebral salt wasting syndrome (CSW), or syndrome of inappropriate secretion of antidiuretic hormone (SIADH) [2]. CDI is usually associated with hypernatremia and polyuria, while the CSW presents with

Fig. 1. A CT cerebral angiography showing occluded basilar artery just above the vertebral arteries union. CT, computed tomography.

Fig. 2. A brain CT showing ischemic infarction involving the cerebellum, brain stem, occipital lobes, temporal poles, and hypothalamic nuclei with a significant cytotoxic edema. CT, computed tomography.

hyponatremia and polyuria. On the other hand, SIADH presents with hyponatremia without intravascular volume depletion [3, 4]. The coexistence of CDI and CSW was described in a few other articles [1, 5, 6]. Yet, to our knowledge, this is the first report of coexisting CDI and CSW after cerebrovascular infarction. The management of electrolytes and fluid imbalance in such patients is usually challenging as considering the neurologic status of the patient and the poor prognosis of the original brain insult.

Case Presentation

A 50-year-old male patient presented to the hospital with a history of dizziness and vomiting twice at home after falling and head trauma. On presentation, the patient was obtunded, confused with dysarthria. Glasgow coma scale (GCS) was 13/15. Pupils were equal and reactive; the patient was not cooperative enough to perform a comprehensive neurologic examination, including the vi-

sion status and visual field. However, tendon and plantar reflexes were normal. He had right parietal skin lacerations and hematoma. No other neurologic deficits were detected. Computed tomography of the brain (brain CT) showed right parietal subgaleal hematoma with no brain parenchymal abnormality. He had a medical history of old cerebrovascular ischemic stroke with no residual effect, hypertension, and polycythemia vera.

A few hours later, the patient developed further deterioration of consciousness. The GCS dropped to 9/15, for which follow-up brain CT was performed.

Findings in the brain CT showed signs of acute ischemic infarctions involving cerebellar and occipital lobes bilaterally. Thrombolysis or intra-arterial thrombectomy was considered and discussed with the neurologist. However, the intervention was contraindicated due to a history of head trauma before the presentation and a history of cerebrovascular ischemia of less than 1 year.

As the GCS was plummeting, the patient was intubated, mechanically ventilated, and shifted to the intensive care unit. Further assessment by CT cerebral angiography revealed occluded basilar artery just above the vertebral arteries union denoting acute posterior circulation infarctions (shown in Fig. 1). The neurologist recommended antiplatelets, statins, and proper hydration.

The next day, neurologic examination showed unequal pupils and loss of gag reflex, so an urgent brain CT was done. Radiological progression of the ischemic infarction was prominent which included the cerebellum, brain stem, occipital lobes, temporal poles, and hypothalamic nuclei. Additionally, significant cytotoxic edema and pontine petechial hemorrhages were noticed (shown in Fig. 2). Hyperosmolar brain dehydration measures were tried; mannitol 20% was administered at a dose of 100 mL every 8 h for 48 h. However, the patient gradually developed bilateral dilated fixed pupils and loss of most of the brain stem reflexes except he was not apneic.

During the hospital stay, the patient developed several attacks of disturbed sodium homeostasis (shown in Table 1). Initially, the patient developed polyuria, serum sodium started climbing up reaching 161 mmol/L and serum osmolality increased to 351 mOsm/kg. Urine sodium level was 188 mmol/L with high urine osmolality 661 mOsm/kg. These findings were believed to be a coexisting CDI and CSW, especially since the patient did not receive high sodium fluid nor hyperosmolar therapy at least 2 days before this event.

However, CDI was the provisional diagnosis. So, desmopressin was administered to manage the significant hyponatremia and the patient showed a good response

Table 1. The range of laboratory findings and polyuria during the episodes of CDI and CSW

	Serum Na, mmol/L	Serum osm. mOsm/kg	Urine Na, mmol/L	Urine osm. mOsm/kg	Polyuria	Diagnosis
First episode	150–161 ↑↑	321–351 ↑↑	107–188 ↑↑	584–661 ↑↑	+	Combined CDI and CSW
Second and third episodes	116–133 ↓↓	246–283 ↓↓	87–129 ↑↑	438–564 ↑↑	+	CSW
Fourth episode	151–165 ↑↑	322–353 ↑↑	20–44 ↓↓	215–262 ↓↓	+	CDI

Na, sodium; osm., osmolality; CSW, cerebral salt wasting syndrome; CDI, central diabetes insipidus.

Fig. 3. A brain CT showing features of hypoxic brain injury and resolving cytotoxic edema. CT, computed tomography.

as regards urine output and correction of hypernatremia within the next few days. Follow-up brain CT revealed features of diffuse hypoxic-ischemic brain injury with resolved cytotoxic edema (shown in Fig. 3).

A few weeks later, the patient developed hyponatremia and serum hypo-osmolality (serum sodium 119 mmol/L and serum osmolality 263 mOsm/kg). Differential diagnosis of CSW versus SIADH was raised. However, as the patient had significant polyuria, the running diagnosis was more of CSW. Therefore, the patient was given fludrocortisone and normal saline infusion till the normalization of serum sodium. A similar episode occurred again after 2 months and was treated likewise.

Three weeks after the last CSW episode, the patient developed polyuria again. This time it was associated with hypernatremia (serum sodium 165 mmol/L), high serum osmolality (353 mOsm/kg), low urine sodium (20 mmol/L), and low urine osmolality (215 mOsm/kg). These results were consistent with CDI that was managed by intravenous desmopressin acetate and optimization of intravascular volume with hypotonic solutions till serum sodium normalized.

Consequently, this patient stayed for more than 10 months, he was in a deep coma with a persistent vegeta-

tive state all through the course in the intensive care unit. He developed several attacks of sepsis, renal and liver impairments. Finally, he succumbed due to septic shock and multi-organ failure.

Discussion

Abnormal sodium homeostasis in critical patients with neurologic disorders could present as CDI, CSW, or SIADH [7]. In this case report, a rare coexistence of CDI and CSW was diagnosed based on the clinical picture and laboratory investigations, including serum sodium, serum osmolality (calculated or measured), urine sodium, and urine osmolality levels. The dramatic response to the prescribed medications confirmed the diagnosis.

The CDI is relatively common. Its overall incidence is about 3.7% among neurosurgical emergencies, and 6.7% in post-craniotomy of pituitary tumor patients [8, 9]. The CDI is usually presented with hypernatremia and polyuria (ranging from 2 up to 10 L/day). The degree of polyuria reflects the level of antidiuretic hormone deficiency [3, 10]. The diagnostic criteria of CDI include hypernatremia >145 mmol/L, urine output >200–250 mL/h, serum osmolality >300 mOsm/kg, and urine osmolality <300 mOsm/kg. Furthermore, a positive response to desmopressin acetate is a deemed confirmation for CDI diagnosis [11]. On the other hand, hyponatremia in critical patients with neurologic disorders could present as SIADH or CSW. Nevertheless, other causes may contribute to hyponatremia such as over administration of hypotonic solutions, adrenal insufficiency, and hypothyroidism. So, it is crucial to define the actual cause of hyponatremia meticulously as the subsequent treatment protocols are different [4]. CSW was first described in the 1950s as an explanation for the occurrence of natriuresis and hyponatremia in patients with cerebral diseases [12]. It occurs in about 7% of patients presented with hyponatremia after subarachnoid hemorrhage [13]. The actual

mechanism of CSW pathology is not clear. However, it may emerge from elevated levels of circulating natriuretic agents as well as disturbed sympathetic innervation to the kidneys which result in increased sodium excretion and contracted intravascular volume. Subsequently, low intravascular volume stimulates baroreceptors to release more antidiuretic hormone and results in hyponatremia [14, 15]. CSW is diagnosed with hyponatremia <135 mmol/L, intravascular volume depletion, and negative sodium balance. Nonetheless, SIADH has a similar laboratory picture, yet no polyuria is present so, the patient is usually euvolemic to hypervolemic if compared to the hypovolemic status with CSW [16]. Differentiating between the 2 diagnoses is of paramount importance as the CSW often responds to normal saline infusion replacement and fludrocortisone therapy. In contrast, SIADH responds better to fluid restriction [17, 18].

In our case, the patient had massive posterior circulation cerebrovascular infarction and several attacks of abnormal sodium hemostasis. The first attack was believed to be a CDI because of polyuria and hypernatremia. Interestingly, the coexistence of CSW was raised due to the concomitant high urine sodium and osmolality with the absence of hyperosmolar therapy nor overloading with sodium-containing intravenous fluids in the preceding 2 days. This episode was managed successfully with desmopressin acetate and hypotonic fluids administration. Apparently in this episode, the CSW was masked by the coexisting CDI. The picture of CSW manifested by hyponatremia and polyuria was more evident in the subsequent 2 episodes. Fludrocortisone and optimization of intravascular volume were the cornerstones of the management of these episodes. Subsequently, the last episode was marked with hypernatremia and polyuria. CDI was the best explanation, especially with low urine sodium and improvement of the condition after desmopressin acetate administration.

Conclusion

The coexistence of CDI and CSW is a rare incidence even in patients with neurologic critical conditions. The original neurologic insult, which is cerebrovascular infarction, makes this case report unique when compared to the original etiologies of other reports. Full consideration of serum electrolytes, hemodynamic state, and urine electrolytes in view of the clinical picture is essential for proper management. Administration of the appropriate medications, for example, desmopressin acetate in CDI and fludrocortisone in CSW, in conjunction with

optimization of the intravascular volume, are the cornerstones of successful management. Unfortunately, due to the rarity of this coexistence, early recognition is hard to be achieved and subsequently proper management. Additionally, the severe original neurologic condition contributes to the bleak outcome of such patients.

Statement of Ethics

Written informed consent was obtained from the patient's son for publication of this case report and any accompanying images. The consent could not be obtained from the patient as he was in a deep coma and then expired. However, as a case report, the ethical approval was not required in accordance with the Dubai Scientific Research Ethics Committee policies.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

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Author Contributions

The authors were part of the treating team, Dr. M.A.E.O. and Dr. H.F.K. had written the case presentation. Dr. H.K. collected the data and extracted the images. Co-authors reviewed the case presentation and data collection. The discussion was collectively written by the full team. Dr. M.A.E.O. and Dr. A.M.H.S. prepared the final copy of the manuscript, which was reviewed and approved by the full team.

Data Availability Statement

All data generated or analyzed during this study are included in this article. Further enquiries can be directed to the corresponding author.

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