Casos clínicos

Cerebral salt wasting syndrome: postoperative complication in tumours of the cerebellopontine angle

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ABSTRACT

Cerebral salt wasting (CSW) is a rare complication in posterior fossa tumour surgery. We present two patients with cerebellopontine angle (CPA) tumours who developed cerebral salt wasting postoperatively. Both patients deteriorated in spite of intensive fluid and salt replacement. On CT scan the patients presented mild to moderate ventricular dilation, which was treated with an external ventricular drainage. After the resolution of hydrocephalus, fluid balance rapidly returned to normal in both patients and the clinical status improved. Identification and treatment of secondary obstructive hydrocephalus may contribute to the management of CSW associated to posterior fossa tumour surgery.

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Palabras clave:
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Síndrome pierde sal cerebral
Hidrocefalia
Hiponatremia
Cirugía

RESUMEN

El síndrome pierde sal cerebral (CSW, en sus siglas en inglés) es una complicación rara en la cirugía de los tumores de la fosa posterior. Presentamos a 2 pacientes con tumores del ángulo pontocerebeloso que desarrollaron un CSW posquirúrgico. Ambos pacientes tuvieron un empeoramiento pese a la fluidoterapia y la reposición de sal intensivas. La tomografía computarizada (TC) mostraba una dilatación ventricular leve a moderada que fue tratada mediante un drenaje ventricular externo. Tras la resolución de la hidrocefalia el balance hidroelectrolítico se normalizó rápidamente en ambos pacientes y su situación clínica mejoró. La identificación y el tratamiento precoz de la hidrocefalia obstructiva pueden contribuir al tratamiento del síndrome pierde sal asociado a la cirugía de tumores de la fosa posterior.

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Introduction

Hyponatremia is the most common electrolyte disorder in hospitalized patients and carries a significant increase in the risk of mortality. It is associated with numerous disorders of the central nervous system like subarachnoid hemorrhage, trauma, infections and tumors. Excluding excessive fluid administration and diuretics, the most frequent form of hyponatremia in the neurosurgical patient consists in hypotonic hyponatremia with elevated natriuresis. This can occur in the context of a syndrome of inappropriate antidiuretic hormone secretion (SIADH) or CSW.

CSW was first described by Peters et al. in 1950 as hyponatremia associated to volume depletion and renal sodium (Na+) wasting without an obvious disturbance in the pituitary-adrenal axis in the setting of various neurological disorders. However following the description of SIADH by Schwartz et al, CSW became viewed as a rare disorder and was frequently misdiagnosed as SIADH. The recognition of the particular clinical features of CSW with its own physiopathology and treatment has led in recent years to give a special consideration to CSW in the neurosurgical practice. Some prospective studies have found CSW even more frequently than SIADH among neurosurgical patients.

We present two rare cases of CSW after removal of a CPA tumor. The differential diagnosis of CSW and SIADH and the specific treatment of CSW is discussed. Special emphasis is given to the early recognition and treatment of hydrocephalus in patients with CSW after posterior fossa surgery.

Case reports

Case 1

A 60-year-old woman with a history of hearing loss on the right side, vertigo and tinnitus was admitted to our department. Audiometry revealed a moderate sensorineural hypoacusis (Gardner-Robertson class III). On magnetic resonance imaging (MRI) an extraaxial mass 2.4 × 2 × 1.6 cm occupying the right CPA with intracanalicular extension and slight compression of the brainstem was found (Fig. 1A). No significant ventricular dilation was present. At our institution vestibular schwannomas below 3 cm of maximal diameter are offered either microsurgery or radiosurgery. Our preferred approach for lesions over 2 cm with significant hearing loss is the translabyrinthine approach. The patient was operated through a translabyrinthine approach with facial nerve monitoring achieving a complete tumor resection. The pathologic diagnosis confirmed a vestibular schwannoma. Postoperatively the patient had a normal neurological examination and no signs of complication on the CT-scan (Fig. 1B). On the fourth postoperative day she presented a progressive decline of consciousness with confused speech and drowsiness (GCS score 13), associated to headache and vomiting. Clinically the patient had signs of dehydration. Laboratory findings showed hyponatremia of 118 mmol/L with plasma osmolarity 237 mOsm/L, urinary Na+ 140 mmol/L and urinary osmolarity 517 mOsm/L. Natriuretic peptides and antidiuretic hormone (ADH) levels are not performed on an emergency basis at our institution, therefore based on clinical and laboratory data a CSW was suspected. Treatment was initiated with volume repletion with isotonic saline and a perfusion of hypertonic saline. Six hours later the clinical status of the patient continued to deteriorate presenting a GCS score of 11. Laboratory values showed no response to initial treatment with plasma Na+ 118 mmol/L, plasma osmolarity 238 mOsm/L, urinary Na+ 142 mmol/L and urinary osmolarity 519 mOsm/L. Invasive monitoring showed a central venous pressure (CVP) of 0 mmHg. A CT-scan revealed moderate hydrocephalus and cortico-subcortical edema. An external ventricular drainage (EVD) was placed in the right frontal horn (Fig. 1C-D). After placement of the ventricular drainage the patient improved rapidly. The laboratory control at 12 hours from onset of the symptoms showed a good response with plasma Na+ 129 mmol/L, plasma osmolarity 267 mOsm/L, urinary Na+ 51 mmol/L and urinary osmolarity 184 mOsm/L. The urinary output during the first 24 hours was 3000 cc with a negative fluid balance of 700 cc. Treatment was continued with volume repletion for 2 days until the patient presented normal fluid balance and Na+ levels with oral fluid intake as needed. The control CT showed small ventricles and no edema. The EVD was removed on day 6. The patient was discharged 16 days after later in good general health.

Figure 1 – (A) Preoperative MRI showing a right sided CPA tumor with intracanalicular extension. (B) Postoperative CT showing complete tumor removal through a translabyrinthine approach. (C) CT on the fourth postoperative day revealing mild ventricular dilation and cortical edema. D) CT after EVD placement with resolution of hydrocephalus.
Case 2

A 72-year-old woman with a history of colorectal carcinoma and a previously treated lung metastasis was admitted to our unit referring headache and vertigo. The patient presented a fair functional status with Karnofsky score of 80 and ECOG score of 1. The MRI revealed an exophytic mass $2.1 \times 2.3 \times 2.6$ cm originating at the left cerebellopontine fissure (Fig. 2A). The tumor was completely removed through a retrosigmoid approach. The pathological findings suggested an adenocarcinoma metastasis. Postoperatively the patient presented slight facial and lower cranial nerve palsy with otherwise good condition. Three days later she suffered a deterioration of consciousness with GCS 13. Electrolyte values showed decreased plasma Na+ 118 mmol/L and osmolality 255 mOsm/L with urinary Na+ 140 mmol/L and osmolality 483 mOsm/L. Emergency CT showed a hematoma occupying the left CPA with compression of the fourth ventricle and hydrocephalus (Fig. 2B-C). The patient was managed in the ICU with CVP monitoring showing initial pressure of 3 mmHg. Neurological deterioration continued in spite of volume repletion with isotonic saline and a hypertonic perfusion. Analysis after 6 hours showed Na+ 120 mmol/L and osmolality 253 mOsm/L. The hematoma was surgically removed and an EVD was placed (Fig. 2D). The clinical and analytical condition progressively improved presenting 12 hours later plasma Na+ 130 mmol/L and osmolality 276 mOsm/L. The EVD was removed 9 days later. The patient was transferred to the neurosurgical ward 10 days later with normal fluid balance.

Discussion

The pathogenic mechanism of SIADH is excessive ADH release leading to renal water resorption and expansion of extracellular fluid volume (ECF). This causes a dilutional hyponatremia in a state where normal renal handling of Na+ is not altered. CSW was suspected to be an independent entity after observation of patients who developed hyponatremia with negative salt balance and ECF volume depletion. Onset of CSW typically occurs within the first ten days following subarachnoid hemorrhage (SAH), trauma, stroke or neurosurgical procedure. The mechanism by which intracranial disease causes renal salt wasting is poorly understood. Disregulation of neural input into the kidney has been postulated. Decreased sympathetic tone could alter the Na+ reabsorption at the proximal tubule and at the same time reduce secretion of renin and aldosterone in response to low plasma volume. Additionally neural natriuretic factors have been found to play a role in CSW. Atrial natriuretic peptide (ANP) and brain natriuretic peptide (BNP) directly inhibit the Na+ reabsorption in the proximal tubule and indirectly increase natriuresis by inhibiting the renin-angiotensin-aldosterone axis. BNP, primarily produced in the cardiac ventricles and the hypothalamus, is considered the most probable natriuretic peptide mediating CSW.

Distinguishing CSW and SIADH can be difficult given the similarity in laboratory values and the common neurological disorders that may cause them. In fact patients with either syndrome present a hypotonic hyponatremia with concentrated urine and increased urinary Na+ (Table 1). Serum uric acid tends to be low in both disorders. Increased fractional excretion of uric acid in CSW is ascribed to impaired Na+ reabsorption in the proximal tubule while in SIADH it is explained by volume expansion. Correction of hyponatremia only corrects uric acid excretion in SIADH. Increased fractional excretion of phosphate is a marker of volume depletion and values above 20% are very suggestive of CSW. In clinical practice the clue to differentiate CSW and SIADH remains the determination of ECF volume. ECF volume is increased in SIADH and decreased in CSW. Evidence of ECF decrease can be obtained from physical examination, weight loss and negative fluid and salt balance. Several authors recommend invasive monitoring of CVP to establish the diagnosis and control treatment. Usefulness of natriuretic peptides and ADH for the differential diagnosis of hyponatremia is not well established. Our two patients showed the typical pattern of hyponatremia caused by CSW: signs of dehydration with low CVP and natriuresis with high urinary output. CSW can be worsened by fluid restriction if the disorder is treated like a SIADH. The treatment of CSW is based on intravascular volume repletion with intravenous saline and Na+ replacement. Mineralocorticoids like fludrocortisone may be used to reduce the loss of Na+.
To our knowledge there is only one report in the literature on tumors of the CPA causing CSW after surgery. Roca-Ribas et al. describe two acoustic neuromas that developed CSW postoperatively. Both our patients were initially treated with fluid and salt replacement but did not show clinical response or analytical improvement during the first 6-12 hours. The CT scan in both cases revealed mild to moderate ventricular dilation and edema. Case 2 also had a postoperative hematoma in the left CPA with moderate mass effect that was surgically removed. Interestingly both patients not only improved neurologically after placement of an external ventricular drainage, but also had a rapid response to CSW treatment thereafter. This observation suggests that hydrocephalus may play a role in the pathogenesis of CSW. Natriuretic peptides have a regulatory role in brain water and electrolyte content and have been found in different experimental animal models to reduce intracranial pressure and brain edema. Serum and cerebrospinal fluid levels of ANP and BNP have been shown to rise with increased intracranial pressure in patients with SAH or traumatic brain injury. Recent data show that ANP down-regulates intracranial pressure in patients with SAH or traumatic brain injury. It is speculated that CSW could be a protective mechanism against intracranial hypertension in neurological diseases such as SAH, trauma or stroke.

### Table 1 – Differential diagnosis of cerebral salt wasting (CSW) and syndrome of inappropriate antidiuretic hormone secretion (SIADH)

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### References