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CASE REPORT

Managing adipsic diabetes insipidus following anterior communicating artery aneurysm in a subtropical climate

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Key Clinical Message

Diabetes insipidus without perception of thirst, as may follow an anterior communicating artery aneurysm, requires prescription of fluid intake as well as desmopressin. The management goal of maintaining a normal serum sodium is rendered more challenging in a humid subtropical environment, where insensible losses are higher.

Keywords

Adipsic diabetes insipidus, anterior communicating artery aneurysm, subarachnoid hemorrhage.

Introduction

Adipsic diabetes insipidus is a rare complication of anterior communicating artery clipping. Maintenance of a normal serum sodium concentration is dependent on both an intact thirst sensation and the action of arginine vasopressin (AVP), also known as antidiuretic hormone. Thirst osmoreceptors and cell bodies containing AVP overlap in location in the hypothalamus and share a common blood supply through branches of the anterior communicating artery [1]. Thus, disruption of this blood supply can result in both lack of thirst and AVP synthesis, in response to appropriate stimuli of hypernatremia and hyperosmolality. Herein, we report the case of adipsic diabetes insipidus following anterior communicating artery clipping. At presentation, the patient had no perception of thirst despite the presence of significant hypernatremia. The fluid management in this case is rendered more complex due to the patient having poor short-term memory and the subtropical climate in Queensland, Australia.

Case History

A 36-year-old woman was transferred to our hospital for further investigation and management of a subarachnoid hemorrhage (SAH). She had initially presented to the Emergency Department following a syncopal episode on a background of a 2-day history of a severe frontal headache associated with nausea and vomiting. Past history included an appendicectomy and tubal ligation. She was a current smoker.

Physical examination at the time of admission revealed weight of 85 kg, heart rate 84/min, blood pressure 140/85, respiratory rate 16 breaths/min, and oxygen saturation 99% on room air. Glasgow coma scale (GCS) was 15. Cardiorespiratory and neurological examination was unremarkable.

Computerized tomographic angiography (CTA) revealed a ruptured 5 mm anterior communicating artery aneurysm (Fig. 1). Treatment was commenced with intravenous fluids and regular nimodipine. She subsequently underwent clipping of the anterior communicating artery aneurysm. Neurological observations revealed a GCS of 13–14 following the procedure. Endocrinology was notified for review after her urine output was recorded at 7.5 L on the day of the procedure.

Differential Diagnosis, Investigations, and Treatment

Preoperative serum sodium concentration on the morning of surgery was normal at 136 mmol/L (reference

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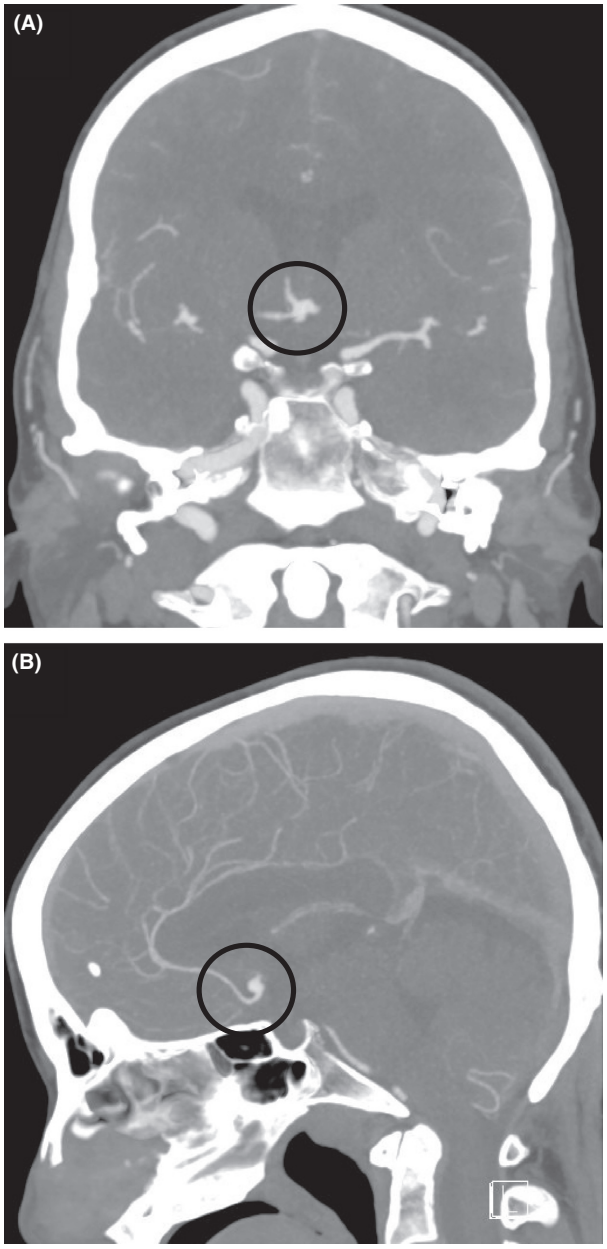

Figure 1. Aneurysm arising from the anterior communicating artery and proximal A2 segment of the right anterior cerebral artery – (A) coronal and (B) sagittal views.

range: 135–145). The following morning, the serum sodium level was 153 mmol/L and serum osmolality was 318 mOsmol/kg (reference range: 280–295). Serum sodium concentration had further increased to 156 mmol/L 2 h later. The urine was dilute, with hourly urine output elevated at 300–400 mL/h.

A clinical diagnosis of central diabetes insipidus was made. Despite the clinical presence of volume depletion and significant hypernatremia, the patient had no perception of thirst. Given the anatomy and location of the aneurysm, the most likely diagnosis was adipsic diabetes insipidus secondary to clipping of the anterior communicating artery aneurysm. Treatment was initiated with 1 μg of subcutaneous desmopressin. Repeat serum sodium concentrations initially fluctuated between 127 and 153 mmol/L during the acute surgical recovery period. Urine osmolality was measured several times while in the Neurosurgical High Dependency Unit. Values varied from 159 to 823 mosmol/kg, depending on the temporal relationship with the administered desmopressin. There was no evidence of major temperature dysregulation – in the immediate postoperative period, temperature varied between 36.5 and 38.1°C, and by the second postoperative week was consistently between 36 and 37°C.

A pituitary hormone profile was performed to exclude anterior pituitary dysfunction. Serum cortisol concentration was 526 nmol/L at 08:10 h, free thyroxine concentration 7.2 pmol/L (reference range: 7–17), thyroid-stimulating hormone (TSH) concentration 0.9 mU/L (reference range: 0.3–4.5), prolactin 374 mU/L (reference range: 71–566), estradiol 141 pmol/L, follicle-stimulating hormone (FSH) 1.8 U/L (reference range: 1–8), and luteinizing hormone (LH) 0.6 U/L (reference range: 1–12).

During the acute period, the patient was initially monitored with hourly urine output, twice daily electrolytes, and daily weighs. Initial parameters for repeat desmopressin administration were the presence of breakthrough polyuria, defined as a urine output of >350 mL/h for two consecutive hours, or >300 mL/h for three consecutive hours. Several doses of parenteral desmopressin were required over the following 72 h. Treatment was then commenced with regular oral desmopressin 100 μg in the morning and 200 μg in the evening. The patient continued to have no perception of thirst. Based on her fluid balance chart after commencing oral desmopressin, the estimated requirements were approximately 2 L of water daily. Therefore, she was placed on a regular water prescription of 250 mL eight times a day. This was administered with close nursing supervision. It was not deemed necessary to perform a formal water deprivation test or infusion of hypertonic saline to confirm the diagnosis in this clinical setting.

She was then transferred to the Brain Injuries Unit for rehabilitation of short-term memory impairment. Serum sodium concentration was monitored at regular intervals and was stable prior to discharge.

Outcome and Follow-up

After initiation of regular oral desmopressin and a fixed dose water prescription, the patient was able to maintain a normal serum sodium concentration in hospital. Serum

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sodium concentration was initially monitored on a daily basis, with the interval being extended to twice weekly prior to discharge. Serum sodium concentration was maintained within the range of 136–144 mmol/L in the 2 weeks prior to discharge. Discharge planning involved ongoing follow-up with weekly bloods for serum sodium concentration with her general practitioner.

Early follow-up at Endocrinology outpatients disclosed mild–moderate hypernatremia (serum Na 145–156 mmol/L). The desmopressin was increased to 200 μg twice daily and the water prescription also modified, aiming for approximately 1 L above urine output. It was thought that insensible losses in the Queensland late spring/summer accounted for the increased fluid requirements, as 24-h urine volumes were not excessive and there was no nocturia. Further review 2 months later revealed some improvement in cognitive function and stable normal electrolytes (serum Na 139–142 mmol/L) on the above regimen. Thirst perception remained absent. The frequency of electrolyte testing was reduced to monthly, 4 months after discharge.

Longitudinal follow-up out to 15 months post SAH has been undertaken. While improved, significant short-term memory impairment persists. Repeat assessment at 1 year indicates that her anterior pituitary function remains intact and she has regular menses. Serum sodium concentrations have remained within the range of 139–147 mmol/L since she has been living with family support in her own home.

Discussion

Adipsic diabetes insipidus is a rare condition. The most commonly reported causes include congenital lesions such as septo-optic dysplasia, germinoma, anterior communicating artery rupture or clipping, and craniopharyngioma [2]. Crowley et al. [3] have reported a series of 13 patients, of whom four were the result of anterior communicating artery aneurysm surgery. A case report and review of the literature published in 2008 revealed 10 cases of adipsic diabetes insipidus following clipping of an anterior communicating artery aneurysm [2]. Two subsequent case reports have since been identified [4, 5].

Even in the presence of hypernatremia, our patient repeatedly denied any perception of thirst. This provides significant long-term management issues in the patient with coexistent short-term memory impairment. Published cases have continued management in the community by using a combination of regular desmopressin and a fixed dose water prescription [6, 7]. Calculating an ideal body weight when the patient is euvolemic provides a set point from which water intake can be adjusted. A behavioral modification technique for the patient with short-term memory impairment has also been described with the use of a wristwatch alarm [8]. This report demonstrated the success of a staged technique, first by regular prompting by staff and initiation of a reward system when all water was consumed in a day. The rewards were gradually withdrawn and the patient maintained a stable serum osmolality by the use of a wristwatch alarm as the prompt to drink.

The present case had isolated diabetes insipidus in the presence of normal anterior pituitary function. Once the immediate postoperative period had passed, a regular twice daily dose of oral desmopressin coupled with a prescribed fluid intake resulted in stable serum sodium concentrations while in the Rehabilitation ward. The challenge was to maintain this level of stability when she is living in the community. This is particularly important given the patient's subtropical geographical location of southeast Queensland, Australia where there are high ambient temperatures and humidity during the summer months (latitude 27° South, mean maximum ambient temperature 28–29°C, humidity 65–70%), which will increase insensible fluid losses. This required adjustment in her water prescription, over and above what would generally be required in a temperate climate.

Patients with adipsic diabetes insipidus have considerable morbidity and mortality [3, 9]. This is in part related to associated conditions of nonendocrine hypothalamic dysfunction, such as obesity, temperature dysregulation, and sleep disorders, but also an apparent increased rate of venous thromboembolism [3] and serious infections [9]. When compared to patients with diabetes insipidus and normal thirst perception, patients with adipsic diabetes insipidus are more than 10 times more likely to develop significant outpatient hypernatremia (>150 mmol/L) [10].

Close monitoring in conjunction with the patient's general practitioner is an essential component of the long-term management. This has resulted in acceptable stability in the patient's serum sodium despite the triple challenge of adipsia, poor short-term memory and humid subtropical climate.

Conflict of Interest

The authors have nothing to disclose.

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