

Adipsic Hypernatremia after Clipping of a Ruptured Aneurysm in the Anterior Communicating Artery: A Case Report

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Adipsia is a rare disorder that occurs due to damage to the osmoreceptor and not feeling thirst despite hyperosmolality. Adipsic hypernatremia can occur when there is damage to the anterior communicating artery that supplies blood to osmoreceptors, and the level of arginine vasopressin secretion varies widely. A 37-year-old woman, suffering from severe headache, was consulted to the nephrology department for hypernatremia and polyuria after clipping of a ruptured aneurysm in the anterior communicating artery. Despite her hypernatremic hyperosmolar state, she denied thirst and did not drink spontaneously. She was diagnosed adipsic hypernatremia by evaluating the osmoregulatory and baroregulatory function tests. Because adipsic hypernatremia is caused by not enough drinking water even for hyperosmolality due to the lack of thirst stimulus, the strategies of treatment are that setting the target body weight when serum osmolality is normal and have the patient drink water until patient reach the target body weight. Adipsic hypernatremia should be considered to be a rare complication of subarachnoid hemorrhage associated with an anterior communicating artery aneurysm.

Key Words: Adipsia, Thirst, Hypernatremia, Anterior communicating artery

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INTRODUCTION

Adipsic hypernatremia which is derived from a defect of the osmotic receptor is a rare disease. When water intake reaction to rising osmolality is decreased, it makes hypernatremia which is called adipsic hypernatremia. In normal cases, serum osmolality is maintained within a narrow range because of arginine vasopressin (AVP) and thirst¹. The amount of AVP release depends on increased serum osmolality and decreased plasma volume. When plasma osmolality increases, the release of AVP is provoked and intake of water is increased due to thirst, which makes osmolality

be constant².

The thirst center is located anatomically close to the osmoreceptor which regulates the release of AVP but separately³. Therefore, the release of AVP is varied in adipsic disease which has a defective sense of thirst. Since experienced a rare case of adipsic hypernatremia associated with loss of thirst activation after anterior communicating artery surgery, we would like to report the diagnosis process, treatment experience, and literature review.

CASE REPORT

A 37-year-old woman was consulted to the nephrology

department due to uncorrected hyponatremia after neurosurgery. Serum sodium was elevated to 154 mEq/L and urine output was increased to 4L per day. Five days before the date of consultation, she was brought to the emergency room because of a shattering headache. She had no specific

medical or familial history. She was a non-smoker and a social drinker who drank once a week. At the emergency room, her blood pressure was 140/80 mmHg, heart rate was 76 beats per min, respiratory rate was 20 breaths per minute, and body temperature was 36.8°C. Her mental status was nearly alert when she arrived at the emergency room. She was diagnosed with subarachnoid hemorrhage with anterior communicating artery aneurysm rupture through brain computed tomography and angiography (Fig. 1). At the emergency room, the blood lab revealed serum sodium of 141 mEq/L, potassium 3.4 mEq/L, and serum osmolality 289 mOsm/kg. Her liver enzyme was normal and kidney function was intact. In a complete blood cell count, hemoglobin 14.1 g/dL, WBC 8,040, and platelet 356,000 were checked. Immediately after neurosurgery, no specific change existed including serum electrolyte except hemoglobin falls to 9.4 g/dL.

In the department of neurosurgery, they used intravenous mannitol for preventing brain edema direct after surgery. On postoperative day (POD) 3, serum sodium concentration and osmolality increased to 156 mEq/L and 337 mOsm/kg, respectively (Table 1). Mannitol and 0.45% saline were infused into intravenous after POD 3. At POD 5, the serum sodium concentration has still remained high at 154 mEq/L and they consulted it to the nephrology department. After following the recommendations for mannitol discontinuation and hypotonic solution administration, serum sodium and osmolality were decreased to 139 mEq/L and 305 mOsm/kg on POD 15, respectively (Table 1). However, urine sodium concentration and osmolality were still low 35 mEq/L

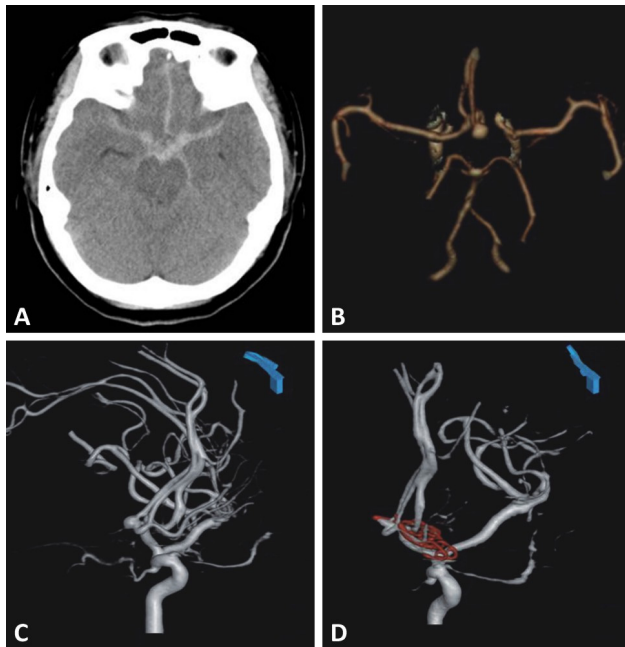


Fig.1. Brain computed tomography (CT) scan at arrival in the emergency room and trans-femoral cerebral angiography (TFCA) findings before and after clipping surgery.

(A) Acute subarachnoid hemorrhage on precontrast brain CT. (B) Aneurysm of anterior communicating artery on brain CT angiography. (C, D) TFCA findings before (C) and after (D) clipping surgery.

Table 1. Changes in serum sodium, and serum and urine osmolality during admission

	POD 0	POD 2	POD 3	POD 5	POD 12	POD 15	POD 24	POD 26	Discharge ↓	Readmission ↓	Outpatient
Serum sodium (mEq/L)	145	150	156	154	173	139	148	138			154
Serum osmolality (mOsm/kg)	292	20	337	331	56	305	314				321
Urine sodium (mEq/L)					46	35	2				26
Urine osmolality (mOsm/kg)					164	132	95				149

←—————→
Desmopressin

POD: postoperative day.

and 132 mOsm/kg, which was thought to be due to an increase in urine output as a result of administering 4-5 liters of fluids per day. As a result of frequent consultation with a nephrologist due to recurrent hypernatremia, there was a possibility of central diabetes insipidus, but a diagnostic test was not possible, they empirically used intranasal desmopressin irregularly. On POD 26, serum sodium was decreased to 138 mEq/L and urine output was decreased by 1.9 liters per day (Table 1). Before discharge, the serum sodium concentration was maintained relatively stable, and she did not complain of any special symptoms. She was discharged without desmopressin spray because the cellar magnetic resonance image taken to confirm the pituitary lesion did not show any specific findings.

Two weeks after discharge, laboratory tests at the nephrology outpatient clinic showed that serum sodium and osmolality increased to 154 mEq/L and 321 mOsm/kg, respectively, and urine sodium and osmolality decreased to 26 mEq/L and 149 mOsm/kg, respectively (Table 1). She did not complain of thirst and polyuria when she visited the nephrology outpatient clinic. Conducting a questionnaire based on laboratory tests showed that the patient rarely drank water because she did not feel thirsty on their own. She was hospitalized for evaluation of a defect of sense of thirst. After admission, massive hydration was done with 5% dextrose solution then serum sodium was decreased

to the normal range. During hydration with 5% dextrose solution, the total input was 3,750 mL/day and the total urine output was 1,800 mL/day, showing no polyuria. The day after admission, she underwent hypertonic saline infusion to confirm the defect of sense of thirst⁴. After discontinued desmopressin and overnight fast, 5% hypertonic saline was infused intravenously at 0.05 mL/kg/min for 2 hours. Then blood was withdrawn for checking serum osmolality and urine osmolality at 30-min intervals during the infusion and thirst via visual analogue scale was estimated at 30-min intervals during the infusion⁵. For AVP measurement, immediately after blood collection, it was sent to the laboratory in an EDTA bottle, centrifuged, frozen at -20°C, and referred to an external consignment agency (GC Labs) to measure it by double anti-body radioimmunoassay method (RK-VPD, Bühlmann, Switzerland). Despite increased serum sodium and osmolality, she did not drink water and did not complain of thirst and urine concentration was inadequate (Fig. 2). When desmopressin was administered after the hypertonic infusion was completed, urine concentration was additionally generated (Fig. 2). Through this test, it was confirmed that she cannot react to hyperosmolality because of the disorder of the thirst center.

The day after the hypertonic saline infusion test, he underwent infusion of nitroprusside for baroregulatory function test. The rate of infusion of nitroprusside was 0.3 mg/kg/min and serum concentration of AVP was measured for every 5 percent of decreased mean blood pressure. Serum concentrations of AVP slightly increased with decreasing mean blood pressure (Fig. 3). A pituitary function test was

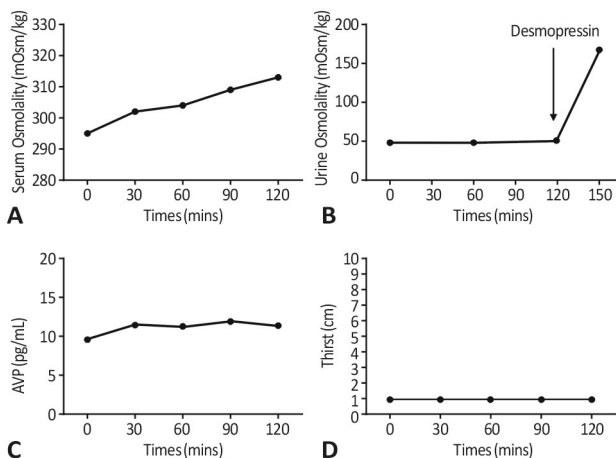


Fig. 2. Result of hypertonic saline infusion test.

Changes in serum osmolality and urine osmolality during hypertonic saline test (A, B). Changes in serum AVP during hypertonic saline test (C). The visual analogue scale of thirst during hypertonic saline test (D).

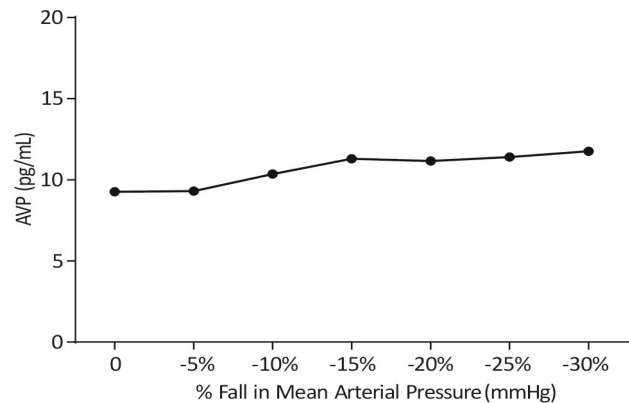


Fig. 3. Result of baroregulatory function test.

Table 2. Result of pituitary function test

	Basal	30 min	60 min	90 min	120 min
GH (ng/mL)	0.04	0.1	7.79	4.07	1.06
Cortisol (ug/dL)	10.86	5.4	11.94	12.4	9.62
TSH (IU/mL)	5.84	36.97	30.41	20.75	13.05
FSH (mIU/mL)	4.98	27.52	36.31	36.86	36.49
LH (mIU/mL)	0.07	6.59	6.44	5.27	4.68
PRL (ng/mL)	12.37	101.53	182.8	86.4	49.32
Glucose (mg/dL)	84	21	81	90	106

GH: growth hormone, TSH: thyroid stimulating hormone, FSH: follicle stimulating hormone, LH: luteinizing hormone, PRL: prolactin.

also performed the next day, to rule out the possible damage to the pituitary gland after brain surgery. Her pituitary function was intact (Table 2). Before the discharge, she received education about routine checks of body weight and the use of desmopressin spray for regulating serum concentration of sodium and plasma osmolality. The body weight was measured at the same time every morning, and if it was lighter than the target body weight (body weight at a serum sodium concentration of 140 mEq/L), water was intentionally ingested, and 10 µg of desmopressin nasal spray was administered 2 times a day.

After discharge, she visited the outpatient clinic of nephrology regularly and was assessed serum concentration of sodium, osmolality, and volume status. During the follow-up period, the patient was maintained in a stable state with only water intake to reach the target body weight. When serum concentration of sodium or osmolality was out of the normal range, we modulated target body weight and serum sodium and osmolality became within normal range. It remained relatively stable during the outpatient follow-up period, but the thirst sense was lost and did not recover for a considerable period of time. Three years after the operation, her sense of thirst was gradually restored, and the euvolemic and eunatremic status was maintained without desmopressin, and now it is completely restored to normal.

DISCUSSION

Plasma osmolality is tightly controlled by osmotic re-

ceptors, regulated within a narrow range, affecting serum sodium concentration. Osmoreceptors maintain osmolality within the normal range by two mechanisms. First, if serum osmolality increases, neural signals derived from osmoreceptors are transferred to the posterior pituitary which is a lesion of AVP release⁶. Activated release of AVP increases water reabsorption in collecting tubules by increasing the number of aquaporin which is a channel for water transport in the membrane of collecting duct cells. Secondly, the osmoreceptor triggers to feel of thirst and intake of free water. Enough intake of free water cause plasma concentration of sodium and osmolality to be normalized.

Postoperative osmotic receptor abnormalities in patients with anterior communicating artery aneurysms are believed to be related to the anatomical regions supplied by the anterior communicating artery. When the hypertonic saline test is performed in this patient, even though osmolality increases, osmoreceptors of these patients can not provoke to feel thirst and secrete additional AVP. In the case of a baroregulatory test that lowers blood pressure in this patient, AVP secretion slightly increases because the reaction in which AVP increases in response to lowering blood pressure is relatively intact consistent with previous reports^{7,8}. In Korea, there have been case reports of patients who do not feel the thirst for hyperosmolality after brain damage with loss of the function of AVP secretion⁹⁻¹².

The difference between this patient's case and most previously reported cases of patients with osmotic receptor dysfunction after anterior communicating artery aneurysm surgery is that the lack of thirst for increased osmotic pressure was similar to that of other patients, but the secretion of AVP was within the normal range. When the hypertonic saline test and the baroregulatory test were performed, the AVP did not additional increase in response to hyperosmolality and low blood pressure. The concentration of serum AVP was maintained similar to the initial concentration. Moreover, at the time of admission to the nephrology department, the daily urine volume was maintained between 1.8 to 2 liters without desmopressin spray. The urine osmolality rose after desmopressin administration in the early phase, but it became stabilized with water intake alone. Thus, we believe that central diabetes insipidus temporarily occurred but was spontaneously resolved.

The main strategy for the treatment of adipsic hypernatremia is to administer an AVP analogue and to encourage intake of sufficient water to prevent dehydration. Periodic evaluation of volume status is a key to managing patients with adipsic hypernatremia. The clinicians should set a target body weight that normalizes plasma concentration of sodium and osmolality and also educate the patients to intake enough water by themselves when their body weight is lower than the target body weight.

Suspicion and recognition of adipsia if hypernatremia develops during treatment for brain lesions is necessary to prevent the development of severe hypernatremic dehydration in the postoperative period. Adipsic hypernatremia should be considered to be a rare complication of subarachnoid hemorrhage associated with an anterior communicating artery aneurysm.

Ethics Statement

This case was approved by the Institutional Review Board of the Gachon University Gil Medical Center (GBIRB2021-428), which waived the need for informed consent.

Conflict of Interest

The authors declare no competing interests.

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