



Adipsic Diabetes Insipidus: A Single-Center Case Series

Forogh Sabzghabaei,^{1,4} Mahsa Akhtar,² Seyed Mahmoud Ramak Hashemi,³ and Reza Mollahoseini⁴

¹Assistant Professor of Nephrology, Department of Medicine, Firoozgar Hospital, Tehran, IR Iran

²General Physician, Department of Medicine, Firoozgar Hospital, Tehran, IR Iran

³Assistant Professor, Department of Neurosurgery, Firoozgar Hospital, Iran University of Medical Sciences, Tehran, IR Iran

⁴Associate Professor, Department of Neurosurgery, Firoozgar Hospital, Iran University of Medical Sciences, Tehran, IR Iran

*Corresponding author: Forogh Sabzghabaei, Assistant Professor of Nephrology, Department of Medicine, Firoozgar Hospital, postal code: 1593748711, Tehran, IR Iran. E-mail: fresh1383@gmail.com

Received 2017 May 29; Revised 2017 August 25; Accepted 2017 November 05.

Abstract

Adipsic diabetes insipidus (ADI) is a rare and challenging complication secondary to several neurosurgical procedures. It can lead to severe hyperosmolality and is associated with high rates of morbidity and mortality. Considering the variable time of thirst sensation recovery, long-term treatment is accompanied with the risk of extreme extracellular osmolality fluctuations. Herein, we report 3 cases of this rare presentation of hypothalamic injury.

Keywords: Adipsia, Diabetes Insipidus

1. Introduction

The occurrence of diabetes insipidus (DI) following neurosurgical procedures for brain tumors may be transient or permanent (1). Adipsia rarely accompanies DI (ADI) due to osmoreceptor destruction. These patients become easily dehydrated and are at a high risk of fluid and electrolyte disorders (2). ADI is often associated with significant hypothalamic dysfunction and complications, such as obesity, sleep apnea, thermoregulatory disorders, seizure, venous thromboembolism, and muscle dysfunction (3-5).

Herein, we present our experiences in the management of 3 rare cases of ADI following neurosurgical procedures.

2. Case Presentation

2.1. Case 1

A 68-year-old female patient underwent total transsphenoidal resection of pituitary adenoma. On the first postoperative day, the urine output was about 9 L with an osmolality equal to 175 mosmol/kg. After 2 days, the sodium (Na) level increased to 160 - 175 mEq/L (Table 1), while blood pressure remained normal; the patient also showed mild tachycardia. Other serum electrolytes, such as potassium, calcium, magnesium, and blood sugar, were in the normal range.

Despite high serum osmolality and full consciousness, the patient showed no desire to drink water. Desmopressin

and fixed water intake were administered with the diagnosis of ADI. After 3 days, the urine output and Na level were found to be in the normal range. In the regular 1-year follow-up, no recovery in thirst sensation was observed, whereas the patient showed a relatively stable serum Na and urine output with desmopressin and fixed water intake.

2.2. Case 2

A 30-year-old female with headache, blurred vision, and diagnosis of suprasellar craniopharyngioma underwent surgery. On the first postoperative day, she was conscious and hemodynamically stable with a urine output of 11 L, Na level of 170 mEq/L (preoperative Na level, 144 mEq/L), potassium level of 3.8 mEq/L, and calcium level of 8.7 mg/dL. She showed no desire to drink water, and treatment was initiated with desmopressin and regular water intake, regardless of adipsia. One week later, the urine output was 2400 ml/day, and Na level decreased to 122 mEq/L. Subsequently, she started developing a sense of thirst. Treatment with desmopressin was ceased, and Na level recovered to normal (Table 1). After 3 years, she presented with permanent DI, requiring desmopressin to the same extent.

2.3. Case 3

A 57-year-old man presenting with severe headache was admitted with a diagnosis of intracranial hemorrhage. Upon admission, urine osmolality was 140 mosmol/kg,

Table 1. The Patients' Characteristics and Laboratory Information

Patient	Pre Operation				Second Post-Operation Day				At Discharge			
	GCS	Serum Sodium, Meq/L	Urine Osmolality mosm,ol/kg	Serum Creatinine, mg/dL	GCS	Serum Sodium, Meq/L	Urine Osmolality, Mosmol/kg	Serum Creatinine, mg/dL	GCS	Serum Sodium, Meq/L	Urine Osmolality, Mosmol/kg	Serum Creatinine, mg/dL
Patient 1 Female 68 years	15	143	980	1.1	15	177	280	0.9	15	144	490	0.9
Patient 2 Female 30 years	15	145	875	0.7	15	159	240	0.8	15	141	385	0.7
Patient 3 Male 57 years	15	130	140	1.2	15	167	235	1.3	15	140	525	1.2

urine volume was 7300 mL/24 h, and serum Na level was 130 mEq/L. On the following day, he underwent surgery due to the aneurysm of anterior communicating artery. On the first postoperative day, Na level was 144 mEq/L and urine volume was normal. On the second day, polyuria appeared and Na level started increasing. On the third day, blood pressure dropped to 90 mmHg, without any evidence of bleeding or fluid loss. The urine volume decreased, and serum Na level increased to 167 mEq/L.

The patient was conscious, but had no thirst sensation. Accordingly, diagnosis of adipsia was confirmed. Treatment with intravenous hyposmolar fluids and pure water was initiated. After 24 hours, blood pressure was 130/80 mmHg, and polyuria with diluted urine appeared (urine osmolality, 240 mosmol/kg). Therapy with desmopressin was started and after 2 days, thirst sensation recovered and DI disappeared.

3. Discussion

ADI is a rare disorder, which is generally secondary to hypothalamic lesions (6). It is attributed to the loss of thirst reflex due to surgical trauma to lamina terminalis, where the osmoreceptor neurons are located (7-9). Limited studies have reported a significantly higher risk of morbidity and mortality in ADI patients (2, 9, 10). Therefore, early diagnosis of this disorder in patients with neurosurgical interventions near the hypophysis or hypothalamus is critical.

In almost all cases, including our patients, diagnosis is based on the evaluation of thirst sensation while the patient is awake and hypernatremic. In these cases, water deprivation test is not required for diagnosis. Increased morbidity may be due to treatable hypothalamic abnormalities, such as obesity, sleep apnea, seizure, and thermoregulatory disorders, which should be examined in patients. One of the limitations of the present study was that we did not evaluate the patients for these disorders.

An interesting point about our patients was the recovery time of thirst sensation. In comparison with case No.

1, who was diagnosed with permanent pituitary adenoma and adipsia, recovery was faster in the other 2 patients. This finding is in contrast to our speculations and previous reports, which suggest that damage to anterior and posterior hypophysis in craniopharyngioma management is more severe than in other surgical interventions causing ADI (1). Management of ADI is generally challenging, as the time of adipsia recovery is variable, and central DI persists despite adipsia recovery. Detection of improvement and treatment cessation are important factors since fatal Na disorders may occur in this period.

In summary, delayed diagnosis of ADI, given the rare nature of this disease, leads to severe complications of the hyperosmolar state (11). Routine adipsia evaluation, combined with water deprivation and vasopressin tests (if necessary), is critical for the diagnosis and treatment of ADI. The fluid intake in patients diagnosed with ADI should be supervised daily, based on the constant volume of oral fluids, daily measurement of fluid balance, body weight, and sodium level (12), especially in patients who are physically unable to care for themselves.

3.1. Conclusions

ADI is a rare and challenging condition to manage. Therefore, careful monitoring and a high index of suspicion are required for its detection. The variable time of recovery in adipsia and DI is the most important issue, especially in the chronic management of patients.

References

- Gonzalez Briceno L, Grill J, Bourdeaut F, Doz F, Beltrand J, Benabbad I, et al. Water and electrolyte disorders at long-term post-treatment follow-up in paediatric patients with suprasellar tumours include unexpected persistent cerebral salt-wasting syndrome. *Horm Res Paediatr.* 2014;**82**(6):364-71. doi: [10.1159/000368401](https://doi.org/10.1159/000368401). [PubMed: [25377653](https://pubmed.ncbi.nlm.nih.gov/25377653/)].
- Arima H, Wakabayashi T, Nagatani T, Fujii M, Hirakawa A, Murase T, et al. Adipsia increases risk of death in patients with central diabetes insipidus. *Endocr J.* 2014;**61**(2):143-8. [PubMed: [24212879](https://pubmed.ncbi.nlm.nih.gov/24212879/)].
- Sabzghabaei F, Rastegar A. Adipsic hypernatremic myopathy. *Iran J Kidney Dis.* 2015;**9**(3):256-8. [PubMed: [25957431](https://pubmed.ncbi.nlm.nih.gov/25957431/)].

4. Smith D, McKenna K, Moore K, Tormey W, Finucane J, Phillips J, et al. Baroregulation of vasopressin release in adipsic diabetes insipidus. *J Clin Endocrinol Metab.* 2002;**87**(10):4564-8. doi: [10.1210/jc.2002-020090](https://doi.org/10.1210/jc.2002-020090).
5. Miljic D, Miljic P, Doknic M, Pekic S, Stojanovic M, Petakov M, et al. Adipsic diabetes insipidus and venous thromboembolism (VTE): recommendations for addressing its hypercoagulability. *Hormones (Athens).* 2014;**13**(3):420-3. doi: [10.14310/horm.2002.1496](https://doi.org/10.14310/horm.2002.1496). [PubMed: [25079469](https://pubmed.ncbi.nlm.nih.gov/25079469/)].
6. Verdin E, Smitz S, Thibaut A, Born J, Legros JJ, Luyckx A. Adipsic hypernatremia in a patient with pseudotumor cerebri and the primary empty sella syndrome. *J Endocrinol Invest.* 1985;**8**(4):369-72. [PubMed: [4067208](https://pubmed.ncbi.nlm.nih.gov/4067208/)].
7. Macias Batista A, Martinez Martin FJ, de Pablos Velasco PL. [Diabetes insipidus and adipsic hypernatremia in a patient with a craniopharyngioma]. *An Med Interna.* 1999;**16**(2):87-8. [PubMed: [10193001](https://pubmed.ncbi.nlm.nih.gov/10193001/)].
8. Collieran K, Sanchez Goettler L, Sloan A. Hypothalamic obesity complicated by adipsic central diabetes insipidus following surgical resection of a craniopharyngioma. *J Clin Hypertens.* 2009;**11**(10):608-10. doi: [10.1111/j.1751-7176.2009.00057.x](https://doi.org/10.1111/j.1751-7176.2009.00057.x).
9. Raghunathan V, Dhaliwal MS, Gupta A, Jevalikar G. From cerebral salt wasting to diabetes insipidus with adipsia, case report of a child with craniopharyngioma. *J Pediatr Endocrinol Metab.* 2015;**28**(3):323-6. doi: [10.1515/jpem-2014-0224](https://doi.org/10.1515/jpem-2014-0224).
10. Zantut-Wittmann DE, Garmes HM, Panzan AD, Lima Mde O, Baptista MT. Severe rhabdomyolysis due to adipsic hypernatremia after craniopharyngioma surgery. *Arq Bras Endocrinol Metabol.* 2007;**51**(7):1175-9. [PubMed: [18157396](https://pubmed.ncbi.nlm.nih.gov/18157396/)].
11. Hu MM, Liu M, Liu W. [Diagnosis and treatment of adipsic diabetes insipidus accompanied with intracranial calcification]. *Zhongguo Yi Xue Ke Xue Yuan Xue Bao.* 2013;**35**(2):161-5. doi: [10.3881/j.issn.1000-503X.2013.02.007](https://doi.org/10.3881/j.issn.1000-503X.2013.02.007). [PubMed: [23643004](https://pubmed.ncbi.nlm.nih.gov/23643004/)].
12. Janus DM, Wojcik M, Zygmunt-Gorska A, Wyrobek L, Urbanik A, Starzyk JB. Adipsic diabetes insipidus in pediatric patients. *Indian J Pediatr.* 2014;**81**(12):1307-14. doi: [10.1007/s12098-014-1421-8](https://doi.org/10.1007/s12098-014-1421-8). [PubMed: [24788913](https://pubmed.ncbi.nlm.nih.gov/24788913/)].