

Primary Polydipsia in Children: Two Case Reports

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Primary polydipsia (PP) is marked by an increase in thirst, and most often presents in patients with psychiatric illnesses. Although uncommon in children, we experienced cases of PP in a 15-month-old boy and a 5-year-old girl. Both were admitted to the hospital with symptoms of polydipsia and polyuria that appeared 1–3 months before admission. Brain magnetic resonance imaging in both patients was normal. A water restriction test was performed after hospitalization and showed normal results. The symptoms improved after the parents were instructed to implement water-intake restriction for 2 weeks. Our report provides useful information for the treatment of PP in children.

Key words: Primary polydipsia, Diabetes insipidus, Children, Water restriction

Introduction

Primary polydipsia (PP) is a clinical disorder characterized by excessive thirst leading to excessive fluid intake and consequential excessive excretion of urine without an obvious cause¹⁾. Polydipsia is defined as the consumption of water higher than 2 L/m²/24 h in children and characterized by a urine volume in excess of 2 L/m²/24 h or 150 mL/kg/24 h at birth, 100–110 mL/kg/24 h until the age of 2 years, and 40–50 mL/kg/24 h in older children and adults^{2,3)}. PP is commonly observed in patients with psychiatric problems such as schizophrenia, it is also known as psychogenic polydipsia¹⁾.

Differential diagnosis based on the symptoms of polydipsia and polyuria is difficult because of the broad associated disease spectrum, including renal, endocrine, and neurological diseases⁴⁾. For example, differentiating PP from diabetes insipidus (DI) can be difficult, and the diagnostic gold standard to understand this difference is a water restriction test. Water restriction tests for older infants and children should be performed in the hospital under close medical supervision. The patient should not be allowed to lose more than 5% of body weight. Monitoring of vital signs (temperature, pulse, and blood pressure), body weight, laboratory tests, urine and plasma osmolarities, and plasma sodium concentration are essential. Each urine void should be recorded and urine volume, specific gravity, and osmolality should be measured. While polyuria in true DI occurs because of defective secretion or response to the antidiuretic hormone (ADH), in PP, there is a disturbance in thirst control that is not related to impaired ADH production or release.

Here, we report the cases of a 15-month-old boy and a 5-year-old girl who

presented with symptoms of PP. As there are few reports on the progression of polydipsia and polyuria in children with PP, the present report will inform the provision of care to PP patients.

Case report

1. Case 1

A 15-month-old boy presented to our clinic with symptoms of polydipsia (150 mL/kg) and polyuria that appeared 1 month prior to his visit. His consumption of water increased such that he woke up 3–4 times nightly to drink water. His thirst was unquenchable. Based on his history, it was apparent that he was born by cesarean section with a birth weight of 3,880 g [50–75 percentile (p)] at 40 weeks and 3 days of gestation age. When he visited our clinic, upon physical examination, his general status was good. His blood pressure was 100/60 mmHg, respiratory rate was 40 breaths/min, and body temperature was 37°C. His body weight was 13 kg (90–95 p), and his height was 77 cm (10–25 p). Upon neurological examination, his muscle tonus and deep tendon reflexes were normal. Other physical examination findings were nonspecific. His laboratory tests were as follows: white blood cell (WBC) count 15,230/ μ L, hemoglobin 13.1 g/dL, platelets 348,000/ mm^3 , sodium 137 mEq/L, potassium 4.8 mEq/L, chloride 109 mEq/L, blood urea nitrogen (BUN) 9 mg/dL, creatinine 0.24 mg/dL, albumin 4.6 g/dL, hemoglobin A1c 5.1%, insulin 3.8 uIU/mL, and C-peptide 1.38 ng/mL. Cortisol level was normal, urine osmolarity was 196 mOsm/kg, and serum osmolarity was 284 mOsm/kg. The spot urine test showed a sodium level 20 mmol/L, potassium level 17.2 mmol/L, and creatinine level 22 mg/dL. The fractional excretion of sodium was 0.2%. A water restriction test was performed after admission and the test result showed that serum osmolarity was unchanged at 277–287 mOsm/kg, while urine osmolarity was elevated at 737 mOsm/kg (Table 1). The ADH level was 6.25 pg/mL at the beginning of the test and 12 pg/mL at the end of the test. Urine output was 551 mL/day. The brain sellar MRI and kidney ultrasonography results were normal. At admission, he was instructed to alter his water-drinking behavior and was provided with psychiatric support. His parents did not allow the child to drink water before sleep

and allowed him to play with toys instead of drinking water. Two weeks later, he was followed up at an outpatient clinic. His body weight was 14.3 kg and daily water intake was 70 mL/kg. Four weeks later, his body weight was 14.2 kg, daily water intake was 35–50 mL/kg, and daily urine output was 40–45 mL/kg.

2. Case 2

A 5-year-old girl presented to our clinic with complaints of 180 mL/kg polydipsia and 150 mL/kg polyuria that appeared 3 months prior to her visit. At that time of the visit, her water intake had increased further, and she had started to wake up 3–4 times every night to drink water. She was born by vaginal delivery and had a birth weight of 3,300 g (50–75 p) at 40 weeks of gestation age. When she visited our clinic, upon physical examination, her general status was satisfactory. Her blood pressure was 95/60 mmHg, respiratory rate was 24 breaths/min, and body temperature was 36.4°C. Her body weight was 22.6 kg (90–95 p), and her height was 114 cm (70–95 p). Upon neurological examination, her muscle tonus and deep tendon reflexes were normal, as were other physical examination findings. Her laboratory tests were as follows: WBC count 7,610/ μ L, hemoglobin 13.6 g/dL, platelets 351,000/ mm^3 , sodium 136 mEq/L, potassium 4.4 mEq/L, chloride 101 mEq/L, BUN 13 mg/dL, creatinine 0.32 mg/dL, albumin 4.9 g/dL, and HbA1c 5.4%. Cortisol levels were normal. Urine osmolarity was 226 mOsm/kg, and serum osmolarity was 280 mOsm/kg. The spot urine test showed a sodium level 11 mmol/L, potassium level 45.8 mmol/L, and creatinine level 21.88 mg/dL. The fraction excretion of sodium was 0.1%. A water restriction test was performed after admission, and her serum osmolarity was consequently unchanged at 283–288 mOsm/kg, while her urine osmolarity was elevated at 1,008 mOsm/kg (Table 2). The ADH level was 5.38 pg/mL

Table 1. Water Restriction Test in a 15-month-old Boy

Time (h)	1	2	3	4	5	6
Body weight (kg)	13.5	13.2	13.1	13	13	13.1
Serum Na (mmol/L)	137	137	137	139	139	139
Serum osmolarity (mOsm/kg)	284	281	287	277	285	287
Urine specific gravity	1.006		1.016		1.023	
Urine osmolarity (mOsm/kg)	196	327	492	-	737	603
ADH (pg/mL)	6.25					12.02
Urine output						1.8 cc/kg/day

at the beginning of the test and 8.06 pg/mL at the end of the test. The brain sellar MRI and kidney ultrasonography results were normal. She altered her water-drinking behavior and was provided with psychiatric support. Her parents were instructed to not allow her to drink water before sleep and daily water intake was limited to 1 L. If she wanted to drink water, it would not be provided immediately, and she would be offered her favorite doll to play with instead. If the interest in water was reduced through play, the amount of water intake could be correspondingly decreased. Two weeks later, follow-up was performed at an outpatient clinic. Her body weight was 23 kg, daily water intake was 40 mL/kg, and daily urine output was 40–45 mL/kg.

Discussion

PP is a specific disorder related to thirst that rarely occurs in childhood. Most PP cases described in the literature concern adults who also have severe psychiatric comorbidities⁵. In patients with PP, symptoms generally develop in three phases. They begin with polydipsia and polyuria, followed by hyponatremia (water is retained as the kidneys fail to excrete excess fluid, resulting in low sodium serum values), and finally water intoxication⁶. Acute hyponatremia develops when excessive fluid consumption leads to a rapid fall of serum sodium. An acute drop in serum sodium level of 10 mEq/L over a few hours may produce recognizable clinical symptoms, including diarrhea, salivation, nausea, ataxia, stupor, and coma. The above three stages did not appear in our patients because the symptoms had recently begun, the kidneys were functioning normally, and the serum sodium levels were normal. In the cases described here, urine concentration normalized when water intake was restricted. PP is most often associated with

behavioral dysregulation of water intake in infancy or early childhood. In children without laboratory signs of diabetes, PP is most often confused with DI. PP can cause physiological suppression of serum ADH, which results in increased output of hypo-osmolar urine². The primary means of distinguishing between DI and PP is the water deprivation test, which is more sensitive than an exogenous ADH challenge.

Consequently, PP is more often confused with central DI than with nephrogenic DI. Several clinical and laboratory findings can aid in the differentiation between these two conditions. In addition, in cases of PP caused by psychogenic factors, drinking water is compulsive; therefore, when water intake is restricted, urine becomes concentrated. Cases of psychogenic PP can be identified by measuring vasopressin concentrations in the absence of any underlying disease if differentiation based on water restriction is insufficient.

Grunberg et al⁷. reported one patient who began to consume excessive water at 20 months of age and was diagnosed with PP at 3 years of age. The authors reported that their patient with PP had recovered 2 months later with the help of water restriction only and without any other treatment. Moreover, that patient underwent a water restriction test for the differentiation between DI and PP⁸. Diagnosis in children with PP is challenging because of difficulties with the water restriction test. The water restriction test is intended to restrict the infant's need to drink water and to monitor urine and blood osmotic concentrations every hour for any changes. It is generally difficult to perform this on an outpatient basis. A urinary osmolarity greater than 600 mOsm/kg during the water restriction test is sufficient for a diagnosis of PP⁹. We described here two patients with PP who were diagnosed via a water restriction test. Additionally, we reported in these cases the progress of polydipsia and its recovery without any pharmacological treatment. The treatment of PP aims to alter the patient's drinking behavior, and fluid restriction and psychiatric support are sufficient for treatment in most cases¹⁰. Restriction of up to 50% to 60% of daily fluid requirements may be required to achieve the goal of inducing a negative water balance⁸.

Our patients were found to have no additional underlying disease based on the water restriction test, brain MRI,

Table 2. Water Restriction Test in a 5-year-old Girl

Time (h)	1	2	3	4	5	6
Body weight (kg)	22.6	22.9	22.6	22.6	22.3	22.3
Serum Na (mmol/L)	137	136	136	136	137	139
Serum osmolarity (mOsm/kg)	283	281	281	283	288	285
Urine specific gravity	1.007		1.016		1.031	
Urine osmolarity (mOsm/kg)	226	325	523	954	1008	921
ADH (pg/mL)	4.28					13.15
Urine output			1.3 cc/kg/day			

or ADH level test. Their symptoms improved within 1 month of observation without any pharmacological treatment. Few studies have reported progress in the treatment of PP patients with water restriction. Our findings suggest that PP treatment in children requires at least 2 weeks of consistent habitual correction via water restriction. The present report provides useful information for the treatment of pediatric PP cases. Notably, our findings suggest that PP treatment in children requires at least two weeks of habitual correction via water restriction.

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Patient consent

This study was approved by the institutional review board (CUH 2018-05-041), and the requirement for informed consent was waived due to the retrospective nature of the study.

Conflicts of interest

No potential conflict of interest relevant to this article was reported.

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