

## CASE REPORT

## Neurology

# An Uncommon Cause of Choreoathetosis

Emma Steffer MS | Nathan Sandalow MD

Rosalind Franklin University of Medicine and Science Chicago Medical School, Chicago, Illinois, USA

**Correspondence**

Nathan Sandalow, Rosalind Franklin University of Medicine and Science Chicago Medical School, Chicago, IL 60064, USA.

Email: [nathan.sandalow@rosalindfranklin.edu](mailto:nathan.sandalow@rosalindfranklin.edu)

**Abstract**

Choreoathetosis is an uncommon presentation in the emergency department setting. The differential diagnosis is broad and includes life-threatening as well as benign causes. Lethal etiologies include metabolic derangements such as hyponatremia as in the case presented here. Hypotonic hyponatremia is the most common electrolyte imbalance and can result from 1 of 2 broad categories of dysregulation: excess free-water intake and solute depletion. Here we describe a case of hypotonic hyponatremia due to a less common route of excess free-water intake. Choreoathetosis as a presenting symptom of hyponatremia is described in case reports. We present a case of a 77-year-old Thai woman who presented to the ED with complaints of weakness, mild headache, confusion, vomiting, and choreoathetoid movements for 1 day. She endorsed chronic, worsening constipation and decreased appetite. She was found to be severely hyponatremic with a serum level of 114 mEq/L requiring admission to the intensive care unit (ICU) for emergent electrolyte correction. She denied any diuretic use or excess oral water or alcohol intake. The etiology was unclear until a careful history was taken, whereupon it was revealed that she had been self-administering tap-water enemas excessively for relief of constipation. Choreoathetosis resolved with careful electrolyte correction. A home-administered tap-water enema leading to hyponatremia and choreoathetosis is a subtle presentation that underscores the importance of careful social history-taking, especially when dealing with vague or non-specific symptoms. We review some more common causes of hyponatremia and discuss its initial management.

**KEYWORDS**

choreoathetosis, cultural history, enema, hyponatremia, sodium homeostasis

## 1 | INTRODUCTION

Choreoathetosis is an uncommon presentation in the emergency department (ED) setting. The differential diagnosis is broad and includes life-threatening as well as benign causes. Lethal etiologies include metabolic derangement such as hyponatremia, as in our cur-

rent case. Choreoathetosis as a presenting symptom of hyponatremia is described in case reports.<sup>1</sup> Hypotonic hyponatremia is the most common electrolyte imbalance and is defined as a serum sodium of less than 135 mEq/L<sup>1,2</sup> along with a serum osmolality of less than 280 mOsm/kg. This can result from 1 of 2 broad categories of dysregulation: an excess of water intake or solute depletion through a variety of mechanisms that are outside of the scope of this paper.

Supervising Editor: Robert Levine, MD

This is an open access article under the terms of the [Creative Commons Attribution-NonCommercial-NoDerivs](https://creativecommons.org/licenses/by-nc-nd/4.0/) License, which permits use and distribution in any medium, provided the original work is properly cited, the use is non-commercial and no modifications or adaptations are made.

© 2023 The Authors. *JACEP Open* published by Wiley Periodicals LLC on behalf of American College of Emergency Physicians.

## 2 | CASE REPORT

A 77-year-old Thai woman was brought to the ED by her daughter with complaints of weakness, confusion, non-bloody non-bilious vomiting, and strange movements for 1 day. She had mild headaches and intermittent vertigo for the last 24 hours. The headache was indolent in onset and diffuse. No household members were ill. She reported no history of diuretic use and denied excess water or alcohol intake. She reported feeling mildly constipated for approximately 4–6 weeks, with decreased appetite. She had been using tap-water enemas chronically for decades and did increase the frequency over the last month because of constipation. She reported that tap water enemas were commonly recommended and used in her native culture. It is recommended to use approximately 1–2 times per month. Her practice was to boil 1200 cc of tap water, allowing it to cool to room temperature before infusion. Over the last month, she had increased enema use from 1–2 times per month to 2–3 times per week. She was born in Thailand and immigrated to the United States in 2009. She had been importing the enema apparatus from Thailand ever since she emigrated.

On arrival in the ED the vital signs are unremarkable. The physical examination is notable for an anxious appearance despite denying subjective feelings of anxiety. She continuously fidgets throughout the examination. Neurologic examination is notable for frequently shifting in the bed along with diffuse non-purposeful movements, primarily of bilateral hands and feet; however, this also involves the arms and legs to a lesser extent. The movements are consistent with choreoathetosis. There is no pill-rolling tremor. There is no intention tremor. The cerebellar examination is limited due to these movements; finger-to-nose testing is limited, as is heel-shin testing. She is able to ambulate with a steady gait.

Labs on arrival demonstrated a serum sodium level of 114 mEq/L, serum osmolality 242 mOsm/kg, serum creatinine 0.6 mg/dL. Urine osmolality was measured at 304 mOsm/kg, urine sodium 47 mEq/L, indicating hyposmolar hyponatremia. The total Na deficit was calculated to be 473 mEq. Computed tomography angiography of the head and neck were unremarkable as was the chest x-ray.

Treatment for severe, symptomatic hyponatremia was initiated as follows: An infusion of 60 cc of 3% saline solution was given over 2 hours via central access and total fluid intake was restricted to 800 mL per day. Repeat Na level measured 6 hours after admission improved to 120 mEq/L and urine osmolality decreased to 206. Desmopressin therapy was initiated to prevent overcorrection and the subsequent risk of osmotic demyelination syndrome (ODS).<sup>3</sup> On repeat physical examination 12 hours after presentation, her symptoms of headache, nausea, vomiting, and choreoathetosis had resolved. Repeat Na at 16 hours was 122 mEq/L, raising concern for overcorrection. 100 cc of 5% dextrose were given to prevent overcorrection. At 24 hours, serum Na remained 122 mEq/L, and at 48 hours it increased to 125 mEq/L. She remained in the hospital for 4 days. Upon discharge, her serum sodium concentration was 135 mEq/L and she had no signs of ODS.

## 3 | DISCUSSION

Choreoathetosis is a movement disorder characterized by abrupt, involuntary movements that often convey a feeling of restlessness to the examiner. It is distinguished from tremors and dystonias by the unpredictable nature of the movements and results from dysregulation of neuronal networks interconnecting the basal ganglia.<sup>4</sup> The differential diagnosis for chorea is protean and includes hereditary and acquired dysfunctions of the basal ganglia. Hereditary causes were deemed very unlikely in this patient, given the acuity and age at onset. Acquired causes include drugs, toxins, metabolic or endocrine derangement, cerebrovascular events, autoimmune syndromes, infections, or neoplasms/paraneoplastic syndromes. Ischemic strokes affecting the basal ganglia are unlikely to cause bilateral symptoms and are associated with hemiballismus more commonly than diffuse chorea.<sup>4</sup> In acute onset choreas such as the case presented here, drugs, toxins, and metabolic derangements are of primary concern.

Hyponatremia is the most common electrolyte imbalance in the emergency department.<sup>5</sup> While there is limited data on the frequency of hyponatremia, one study cited that in an emergency department in Taiwan, the prevalence of hyponatremia was 3.8%.<sup>6</sup> Another study examining the general population in the United States found that hyponatremia prevalence was approximately 1.7%.<sup>5</sup>

Hyponatremia is classified as acute or chronic and can be caused by physiological abnormalities such as the syndrome of inappropriate antidiuretic hormone secretion, renal failure, heart failure, cirrhosis, or excessive water intake (Figure 1).

Symptoms classically include headaches, delirium, vomiting, seizures, coma, neurogenic pulmonary edema, or cerebral edema.<sup>7</sup> Neurological symptoms are usually seen only when sodium levels drop below 120 mEq/L, indicating severe hyponatremia.<sup>8</sup> In some cases, severe hyponatremia can cause choreoathetoid movements,<sup>9</sup> although this is not a common symptom.

Hyponatremia that arises in less than 48 hours is defined as acute and is most often due to parenteral fluid administration postoperatively or self-induced water intoxication in syndromes, such as psychogenic polydipsia or beer potomania.<sup>10,11</sup> Mild hyponatremia is defined as sodium concentration from 130 to 134 mEq/L, moderate is defined as 120–129 mEq/L, and severe hyponatremia is defined as <120 mEq/L. The goals of treatment of hyponatremia are prevention of any further decrease in sodium levels, alleviation of symptoms relating to hyponatremia, and prevention of cerebral edema. Caution must be exercised, however, to avoid overcorrection and its dreaded complication, central pontine myelinolysis. The rate of correction should be between 4 and 6 mEq Na over 24 hours.<sup>12</sup> Although fluid restriction may be sufficient in some cases, patients such as ours who exhibit neurologic sequelae in the form of headaches, nausea, vomiting, and choreoathetosis, require emergent treatment with 3% hypertonic saline. In patients with chronic hyponatremia due to underlying pathology or ongoing polydipsia, fluid restriction to 50%–60% of usual daily recommended intake is warranted.<sup>13</sup>

### **Physical exam for fluid overload:**

#### **Hypervolemic hyponatremia:**

Urine Na <20: CHF, cirrhosis, nephrotic syndrome

Urine Na >20: renal failure

#### **Euvolemic/hypovolemic hyponatremia:**

##### **Serum osmolality:**

High (>285mOsm/kg): hyperglycemia, mannitol recent IV contrast

Normal (280-285 mOsm/kg): hypertriglyceridemia, hyperproteinemia

Low (<280 mOsm/kg):

##### **Urine osmolality:**

Low (<100 mOsm/kg): GI losses, burns, diuretics, excess free water

High (>100 mOsm/kg): SIADH, hypothyroidism, adrenal insufficiency

**FIGURE 1** Classification of hyponatremia. CHF, congestive heart failure; GI, gastrointestinal; IV, intravenous; SIADH, syndrome of inappropriate antidiuretic hormone secretion.

In this case, the likely etiology of the hyponatremia was the patient's overuse of tap-water enemas for constipation. Traditionally, first-line treatment of constipation includes dietary counseling to increase fiber intake and oral hydration. If increasing fiber is ineffective, bulk-forming laxatives, stool softeners, osmotic agents, and stimulant laxatives are second-line treatments. Enemas are typically reserved for constipation refractory to these interventions. Fleet enemas are a commonly used over-the-counter brand composed of a 19 g of monobasic sodium phosphate monohydrate and 7 g dibasic sodium phosphate heptahydrate per 118 mL dose (per the manufacturer package insert).<sup>14</sup> Mineral oil is a recommended enema substance; however, many at-home enema kits are available with instructions to use clean water, saline, or coffee.

This case report highlights the cultural and historical importance of enemas. Across the globe, enemas have been used for both laxative, cleansing, and ritualistic purposes for thousands of years.<sup>15</sup> Although there are few data on use across cultures, enemas are widely used as treatments for constipation. If a careful history was not taken with this patient, we may never have discovered the cause of the hyponatremia, to devastating consequences. We suspect that cases of hyponatremia secondary to the overuse of enemas are likely underreported. Clinicians should be aware that this is a possible cause of hypotonic hyponatremia and inquire about enema use and other uncommon practices in cases of unclear etiology.

### **3.1 | Relevance to the emergency medicine physician**

Choreoathetoid movements are a rare presenting complaint in the ED. The differential diagnosis includes hyponatremia, which can be caused by excessive fluid intake that may be the result of overuse of hypotonic enemas. Enemas are a common treatment of constipation

and are generally quite safe if used appropriately.<sup>16</sup> This case highlights the importance of performing a detailed history and specifically asking about alternative treatments beyond prescribed or over-the-counter medications. Although there have been reports of colonic irrigation causing hyponatremia,<sup>16</sup> the development of severe hyponatremia causing choreoathetoid movements, however, are less common. Treatment should be aimed at diligently reversing the sodium imbalance while avoiding overcorrection so as to minimize risk of osmotic demyelination syndrome.

### **CONFLICT OF INTEREST**

The authors declare no financial conflicts of interest to disclose.

### **REFERENCES**

- Adrogué HJ, Madias NE. Hyponatremia. *N Engl J Med*. 2000;342(21):1581-1589. doi:10.1056/NEJM200005253422107
- Schmidt B. Die häufigsten Elektrolytstörungen in der Notaufnahme. [The most frequent electrolyte disorders in the emergency department: what must be done immediately?]. *Internist*. 2015;56:753-759. doi:10.1007/s00108-015-3670-7
- Sood L, Sterns RH, Hix JK, Silver SM, Chen L. Hypertonic saline and desmopressin: a simple strategy for safe correction of severe hyponatremia. *Am J Kidney Dis*. 2013;61(4):571-578. doi:10.1053/j.ajkd.2012.11.032
- Cardoso F, Seppi K, Mair KJ, et al. Seminar on choreas. *Lancet Neurol*. 2006;5(7):589-602. doi:10.1016/S1474-4422(06)70494-X
- Mohan S, Gu S, Parikh A, Radhakrishnan J. Prevalence of hyponatremia and association with mortality: results from NHANES. *Am J Med*. 2013;126(12):1127-1137. doi:10.1016/j.amjmed.2013.07.021
- Lee CT, Guo HR, Chen JB. Hyponatremia in the emergency department. *Am J Emerg Med*. 2000;18(3):264-268. doi:10.1016/s0735-6757(00)90118-9
- Sterns RH. Disorders of plasma sodium—causes, consequences, and correction. *N Engl J Med*. 2015;372(1):55-65. doi:10.1056/NEJMra1404489

8. Tandukar S, Rondon-Berrios H. Treatment of severe symptomatic hyponatremia. *Physiol Rep*. 2019;7(21):e14265. doi:10.14814/phy2.14265
9. Nagaratnam N, Icao E, Peric H. Abnormal movements associated with severe hyponatraemia. *Postgrad Med J*. 1997;73(862):503-504. doi:10.1136/pgmj.73.862.503
10. Sterns R. Causes of Hypotonic Hyponatremia in Adults. UpToDate. 2020. Accessed August 11, 2021. from <https://www.uptodate.com/contents/causes-of-hypotonic-hyponatremia-in-adults>
11. Lodhi MU, Saleem TS, Kuzel AR, et al. "Beer Potomania" - A syndrome of severe hyponatremia with unique pathophysiology: case studies and literature review. *Cureus*. 2017;9(12):e2000. doi:10.7759/cureus.2000
12. Sterns RH, Nigwekar SU, Hix JK. The treatment of hyponatremia. *Semin Nephrol*. 2009;29(3):282-299. doi:10.1016/j.semnephrol.2009.03.002
13. Braun MM, Barstow CH, Pyzocha NJ. Diagnosis and management of sodium disorders: hyponatremia and hypernatremia. *Am Fam Physician*. 2015;91(5):299-307.
14. Fleet® Enema Saline, (2021). Accessed from <https://www.fleetlabs.com/constipation-relief-products/enemas/fleet-saline-enema>
15. Doyle D. Per rectum: a history of enemata. *J R Coll Physicians Edinb*. 2005;35(4):367-370.
16. Norlela S, Izham C, Khalid BA. Colonic irrigation-induced hyponatremia. *Malays J Pathol*. 2004;26(2):117-118. [http://www.mjpath.org.my/past\\_issue/MJP2004.2/06-colonic.pdf](http://www.mjpath.org.my/past_issue/MJP2004.2/06-colonic.pdf)

**How to cite this article:** Steffer E, Sandalow N. An Uncommon Cause of Choreoathetosis. *JACEP Open*. 2023;4:e12891. <https://doi.org/10.1002/emp2.12891>