CASE REPORT

Secondary parkinsonism in a patient of psychogenic polydipsia

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SUMMARY

A 44-year-old man presented with history of slurring of speech, slowness in activities, abnormal posturing of the upper limbs and drooling of saliva from the mouth. He had a 5-yearlong history of compulsive water drinking, consuming 12–15 L of water every day. He was earlier evaluated for the same and found to have low serum sodium (126 mEq/L). Presently, he was admitted at a primary care facility with fever and altered sensorium. He was found to have serum sodium of 94 mEq/L. His sensorium improved with hypertonic saline infusion; however, after a lucid interval of 48 hours, he developed florid extrapyramidal symptoms. Contrast-enhanced MRI brain revealed bilaterally symmetric T2-weighted fluid-attenuated inversion recovery hyperintensity in caudate and lentiform nuclei. A diagnosis of extrapontine myelinolysis with secondary parkinsonism was made. He improved significantly with levodopa therapy. Psychogenic polydipsia is an important cause of hyponatraemia. Overzealous correction of hyponatraemia can be counterproductive.

BACKGROUND

Psychogenic polydipsia or compulsive water drinking is often associated with chronic psychiatric disorders. It can lead to considerable morbidity and mortality,¹ much of which is attributed to hyponatraemia. Hyponatraemia usually results when they continue to drink water beyond the point of maximum urine dilution (100 mOsm/kg) and peak antidiuretic hormone suppression.² Demyelination of pontine and extrapontine structures can occur in individuals with hyponatraemia, or more commonly, following overzealous correction of hyponatraemia. Here, we report a case of psychogenic polydipsia presenting with features of parkinsonism. He had hyponatraemia secondary to compulsive water drinking, which was probably exacerbated by acute dengue fever. Rapid correction of serum sodium led to osmotic demyelination in bilateral striatum resulting in classical extrapyramidal symptoms. He improved dramatically with levodopa therapy.

CASE PRESENTATION

A 44-year-old man presented to our outpatient department with history of slurring of speech, slowness in activities, abnormal posturing of the upper limbs and drooling of saliva from the mouth. He was referred from a primary care facility where he had presented a week earlier with fever and altered sensorium. Preliminary

evaluation at the above centre revealed a serum sodium of 94 mEq/L, low platelet count (26×10^9 /L) and normal non-contrast CT scan of the head. ELISA for dengue IgM antibody was positive, explaining the thrombocytopaenia. Lumbar puncture was done and cerebrospinal fluid examination was unremarkable. Attributing altered sensorium to hyponatraemia, he was started on hypertonic saline (3% saline) infusion. Subsequent serum sodium values were as follows: on day 1, 108 mEq/L; on day 2, 120 mEq/L; on day 3, 132 mEq/L and on day, 4–146 mEq/L. His sensorium improved significantly on day 2 of saline infusion. On day 3, he was accepting orally and was communicating properly with his relatives. However, hypertonic saline infusion was continued till day 4. He remained asymptomatic till day 5 and had started to ambulate on his own; however, on day 6, patient developed sudden-onset dystonic posturing of the upper limbs. On day 7, he developed dysarthria, dysphagia and drooling of saliva from the mouth. This was associated with significant clumsiness in his routine activities and he had spent the whole day bound to the bed. He was subsequently referred to our institute.

He had a significant past medical history. Five years ago, in the year 2011, after the unexpected demise of his elder brother, he had resorted to compulsive water drinking. He would complain of unquenchable thirst, compelling him to drink around 12–15 L of water every day and would pass urine amounting to 8–10 L/day. He was evaluated for the same by a family physician in July 2016 when he was found to have hyponatraemia (serum sodium 126 mEq/L). He was clinically diagnosed as having psychogenic polydipsia and was advised for cognitive-behavioural therapy. However, he did not follow up with the physician and continued his compulsive drinking behaviour. He was also addicted to alcohol and would consume 250 g of liquor every week. He was a non-smoker and had no addictions to other illicit drugs. There was no prior history of intake of any antipsychotic or anti-emetic medications. He never had had encephalitis, meningitis or seizures.

On physical examination, he had a masked facies with drooling of saliva from the angles of the mouth. His speech was hypophonic and dysarthric. Objective power testing did not reveal any muscle weakness. However, there was cogwheel rigidity and dystonic posturing of the upper limbs. Bradykinesia and resting tremors were present. He

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Figure 1 Symmetric T2/FLAIR hyperintensities in bilateral striatum (marked in arrows). T2/FLAIR, T2-weighted fluid-attenuated inversion recovery.

had a festinant gait. His sensorium and cognition was intact. Rest of the systemic examination was normal.

INVESTIGATIONS

Investigations revealed haemoglobin level of 12.8 g/dL (12–16 g/dL), total leucocyte count of $9.6 \times 10^9/L$ ($4-11 \times 10^9/L$) and platelet count of $324 \times 10^9/L$ ($150-400 \times 10^9/L$). His serum sodium was 124 mEq/L (135–145 mEq/L) and potassium was 3.9 mEq/L (3.5–5.0 mEq/L). Renal and liver function tests were normal. His fasting blood glucose was 91 mg/dL and corrected serum calcium level of 9.2 mg/dL (8.6–10.2 mg/dL). His serum and urine osmolalities were low (269 mOsm/kg (275–285 mOsm/kg) and 69 mOsm/kg (300–1200 mOsm/kg), respectively). Water deprivation test showed increasing urine osmolality with a maximum of 168 mOsm/kg at the 8th hour. His thyroid function test was normal ($T_3=1.28$ ng/mL (0.8–2.0 ng/mL), $T_4=8.4$ μ g/dL (4.8–12.7 μ g/dL), thyroid-stimulating hormone=3.4 μ IU/mL (0.27–4.2 μ IU/mL). Serum cortisol was low (154 nmol/L, range 171–536 nmol/L), however was appropriately stimuable with 1 μ g synacthen (reaching a peak of 565 nmol/L). Dengue IgM antibody ELISA was positive.

Contrast-enhanced MRI of the brain showed bilaterally symmetrical T2-weighted fluid-attenuated inversion recovery (T2/FLAIR) hyperintensities in the caudate and lentiform nuclei without diffusion restriction and enhancement (figure 1). As the clinical picture was consistent with extrapyramidal symptoms, the possibility of parkinsonism secondary to osmotic demyelination of the basal ganglia was kept.

DIFFERENTIAL DIAGNOSIS

The patient had presented with acute-onset extrapyramidal symptoms following rapid correction of hyponatraemia. In such a clinical scenario, the first differential diagnosis that should be

considered is osmotic demyelination of the basal ganglia and subsequent secondary parkinsonism. Clinically, however, one has to bear in mind other secondary causes of parkinsonism like encephalitis, drugs, toxins and ischaemic insult. The sequence of events and the acuity of symptoms make idiopathic parkinsonism very unlikely.

TREATMENT

Patient was started on levodopa under observation in the ward. Levodopa was started at a dose of 100 mg/day and uptitrated to a maximum dose of 400 mg/day over a period of 2 weeks. He underwent psychotherapy and was advised to restrict water intake to no more than 3 L/day. Selective-serotonin reuptake inhibitors were not started in view of hyponatraemia.

OUTCOME AND FOLLOW-UP

Extrapyramidal symptoms improved significantly with levodopa therapy. There was marked reduction in bradykinesia even with lowest dose of levodopa. As the dose was hiked up to 400 mg/day, tremors and dystonia also disappeared. Dysarthria and drooling of saliva, however, persisted and there was only partial improvement. With fluid restriction, his serum sodium had improved to 136 mEq/L at the time of discharge.

One month later, he was followed up at the outpatient clinic. He had completely recovered by that time. Hence, levodopa was gradually tapered off over a period of another 2 weeks and was monitored for the recurrence of symptoms. However, even after stopping levodopa, he remained asymptomatic. Presently, he is off all medications, doing well and is undergoing psychotherapy sessions. Repeat MRI brain done 1 month after stopping levodopa showed reduced T2/FLAIR hyperintensities in the striatum.

DISCUSSION

Osmotic demyelination syndrome (ODS) was first described by Victor and Adams in 1959. They reported four patients presenting with quadriparesis and pseudobulbar palsy and named the entity as ‘pontine myelinolysis’.³ Surprisingly, all the four patients were alcoholics and the syndrome was attributed to be a sequel of chronic alcoholism. Later, in 1962, it was recognised that similar pathology can occur in areas of brain other than the pons, the so-called ‘extrapontine myelinolysis’ (EPM). However, it was not until 1981 that Kleinschmidt-DeMasters and Norenberg demonstrated a link between ODS and the overzealous correction of hyponatraemia.

ODS is a general term used to describe a constellation of neurological manifestations that develop in the setting of acute osmotic changes, typically with rapid and inappropriate correction of hyponatraemia. It accounts for 0.4%–0.56% of all admissions to neurology services at tertiary care facilities.⁴ Asymptomatic cases are not uncommon and the largest autopsy series have reported a prevalence of 0.25%–0.5% in a general population.⁵ However, certain populations like alcoholics, malnourished and liver transplant recipients are more prone to develop ODS. The postmortem rate of ODS is about 10% in liver transplant patients.⁶

Hyponatraemia causes influx of extracellular water into the neurons leading to cerebral oedema. However, with sustained hyponatraemia, the glial cells and the neurons extrude inorganic and organic osmolytes out of the cells, thereby rendering the brain parenchyma and interstitium hypo-osmotic. This brings down the cellular volume towards normal. With correction of hyponatraemia, fluid moves out of the glial cells and consequently they now try to prevent fluid loss by increasing

production of osmolytes. However, with rapid correction of serum sodium, this process cannot keep pace with the loss of water from the cells. It leads to shrinkage of the cells, damage of the tight junctions and consequent disruption of the blood–brain barrier. This, in turn, provides free access to circulating serum complements, cytokines and other inflammatory mediators into the central nervous system, leading directly to demyelination. Histopathologically, ODS is characterised by well-demarcated areas of demyelination, relative sparing of axons and lack of inflammatory infiltrate in these lesions.⁷

Osmotic demyelination commonly affects the pons, a condition described as ‘central pontine myelinolysis’. Involvement of structures other than the pons is referred to as ‘EPM’. Isolated EPM accounts for no more than 10% of all cases of ODS. Commonly described sites of EPM are the basal ganglia, thalamus, midbrain, hippocampus, cerebellum and deep periventricular white matter. Of late, involvement of the motor cortex, uncus and geniculate body has also been shown.⁸ Classically, patients with central pontine myelinolysis have a biphasic course of illness. With correction of hyponatraemia, sensorium improves; however, between 7 and 14 days after the acute osmotic insult, patient again develops encephalopathy associated with quadriparesis and pseudobulbar symptoms.

EPM affecting the striatum can lead to extrapyramidal symptoms. The incidence of parkinsonism in patients with ODS has been found to be as high as 44%–50%.⁹ Parkinsonism in ODS results from relative dopamine deficiency secondary to reduced presynaptic striatal dopamine transporters.¹⁰ Dopaminergic therapy, principally levodopa leads to symptomatic improvement.¹¹ Prognosis is good, and more than 50% of the patients recover completely.¹² Patients with delayed progression of extrapyramidal symptoms tend to have a poor neurological outcome.¹³

Our index case is an example of secondary parkinsonism resulting from osmotic demyelination of bilateral striatum. However, the circumstances leading to it are somewhat unique. Our patient had psychogenic polydipsia and had a low baseline serum sodium (126 mEq/L) even prior to the acute event. The sudden dip in serum sodium to 94 mEq/L was probably precipitated by the concomitant dengue fever, as hyponatraemia is a recognised complication of this viral illness.¹⁴

The rapid rise in serum sodium, with an absolute increase of 26 mEq/L over a period of 48 hours led to oligodendrocyte apoptosis and loss of myelin. Why the striatum was specifically involved in our patient remains unanswered. The biphasic clinical course of the patient, characterised by an initial improvement in sensorium followed by the development of extrapyramidal symptoms is classical of ODS. The MRI finding of symmetric T2/FLAIR hyperintensities in bilateral striatum is also the characteristic of ODS. The response to treatment with levodopa was marked as has been described in the literature.

Learning points

- ▶ Psychogenic polydipsia can lead to clinically significant hyponatraemia.
- ▶ Overzealous correction of hyponatraemia can lead to osmotic demyelination, affecting the central pons and/or extrapontine subcortical grey matter.
- ▶ Extrapyramidal symptoms can develop secondary to osmotic demyelination of the striatum.
- ▶ Symptoms usually resolve with time; however, residual neurological deficits may persist.

Contributors RP is the primary author. RP assembled the data and drafted the final copy of the paper. LD discussed the case with the radiologists and the psychiatrists. PD coordinated with the neurologists and helped in managing the patient at each and every step. AB provided overall guidance.

Competing interests None declared.

Patient consent Obtained.

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