Case Report

AFFECTIVE SYMPTOMS IN AN INDIVIDUAL WITH CENTRAL PONTINE MYELINOLYSIS – A CASE REPORT

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ABSTRACT

Background: Central pontine myelinolysis is a condition characterized by damage to regions of the brain, the most common site being pontine white matter tracts, following a rapid correction of metabolic disturbances such as hyponatremia. Extrapontine sites that may be involved include the caudate and the lentiform nuclei, putamen, thalami, cerebellum, hippocampus, and cerebral cortex. **Case report:** A female aged 34 years presented with gastritis. Baseline investigations did not suggest an infective picture. Her serum sodium levels were noted as 101 meq/l at admission. Corrective measures were carried out, and serum sodium levels increased to 133 meq/l in 3 days. Within three days, she developed manic symptoms. Psychiatry consultation was sought, and she was admitted to the Psychiatry ward. MRI of the Brain confirmed both central and extrapontine myelinolysis. She was treated with mood stabilizers and antipsychotics and was discharged in two weeks with complete remission of symptoms. **Discussion:** Neuropsychiatric symptoms developing in the background of rapid correction of serum sodium levels have been reported in a few studies previously. Focus on brain areas involved in such cases may provide insight regarding the possible role of these brain areas in the etiopathogenesis of these symptoms.

Key words: Central pontine myelinolysis, hyponatremia, affective symptoms

INTRODUCTION

Central pontine myelinolysis (CPM) is a syndrome characterized by rapid destruction of myelin sheaths of mainly oligodendritic cells.[1] It was first described as osmotic demyelination syndrome (ODS) in alcoholic patients by Adams and Victor in 1959. On pathophysiological considerations, and as an increasing number of manifestation sites in addition to the pons were detected, central pontine myelinolysis (CPM) and extrapontine myelinolysis (EPM) were combined into "osmotic demyelination syndrome." In most cases, ODS occurs in patients with chronic hypotonic hyponatremia, which is corrected too quickly. Known risk factors include severe hyponatremia, alcoholism, thiazide use, hypokalemia, and malnourishment.[1]

Hyponatremia is common in hospitalized patients.[2] Overcorrection of serum sodium level in symptomatic chronic hyponatremia is the major risk factor of ODS development.[3] In hyponatremia, excessive free water in extracellular space moves into the brain cells, causing edema. After some time, the brain cell volume returns to normal by expelling intracellular solutes and water. Overcorrection

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of the serum sodium level increases serum osmolality excessively to induce an out-shift of intracellular water ultimately. This can result in cellular shrinkage during hyponatremia management. This process is the primary pathogenesis of osmotic demyelination syndrome.[3] In the Efficacy and Safety of Rapid Intermittent Correction Compared With Slow Continuous Correction With Hypertonic Saline in Patients With Moderately Severe or Severe Symptomatic Severe Hyponatremia (SALSA) trial, overcorrection was defined as an increase in serum sodium level by >12 mEq/L or >18 mEq/L within the first 24 or 48 hours, respectively.[2]

Early clinical presentation of CPM includes confusion, flaccid quadriparesis (secondary to corticospinal tract involvement), which later becomes spastic), and pseudobulbar palsy, which occur 2-6 days after a rise in serum osmolality. Other observed symptoms are dysarthria, and dysphagia (secondary to corticobulbar fiber involvement), from the involvement of basis pontis; pupillary and oculomotor abnormalities may occur if the lesion extends into the tegmentum of pons. A large lesion in the pontine side can present as a change in consciousness level reflecting the "locked-in syndrome".[1] Encephalopathies, characterized by vigilance disorders, qualitative impairment of consciousness, delirium, and disorders of drive, memory, and concentration, have also been reported. The clinical course can also be oligosymptomatic or asymptomatic.[4]

Extrapontine lesions can result in dystonia, myoclonus, rigor, akinesia, and tremor if basal ganglia is involved.[4] Cognitive symptoms include frontal dysfunctions (e.g., involving problem-solving, planning, drive, impulse control, and emotional control), and concentration disorders; psychiatric symptoms such as depressive or manic syndromes, emotional lability, catatonia, and mutism are also reported.[5]

The advised rate of correcting chronic hyponatremia, especially in high-risk patients, is 4-6 mmol/l for any 24-hour period with a recommended maximum of 8 mmol/l to prevent ODS.[6]

CASE REPORT

A 34-year-old married female was admitted to the Medicine ward with a history of multiple episodes of vomiting over four days duration. She had been on regular treatment for systemic hypertension, which was adequately under control. She was treated with a diagnosis of gastritis. Baseline investigations did not suggest an infective picture. Blood routine, renal function tests, and liver function tests were unremarkable. Her serum sodium level was found to be 101meq/l at admission.

Corrective measures were carried out with 3% hypertonic saline IV infusion, and serum sodium levels increased to 133 meq/l in 3 days. Following this, she developed disorientation to place and disturbed sleep at night with increased psychomotor activity. Gradually, she started exhibiting an elated mood, increased speech, increased level of activities, and overfamiliarity. She had no past history or family history suggestive of neuropsychiatric illness. Psychiatry consultation was sought, and she was started on mood stabilizers and antipsychotics in view of the prominent affective symptoms. CT Brain showed dilated lateral, third, and fourth ventricles. She was symptomatically better and was discharged from Medicine ward with the advice to have follow-up in Psychiatry OPD.

Post-discharge, her affective symptoms worsened, and she began expressing expansive ideas, increased planning, grandiose delusions, and emotional lability; her cognitive functions were intact. Within ten days of discharge, she was readmitted under Psychiatry. Along with mood symptoms, she presented with a headache and had an unsteady gait, which recovered in two days. The possibility of normal pressure hydrocephalus, intracranial space-occupying lesion, electrolyte

Table 1. Timeline of the case

26/06/22	Admitted under the Department of Medicine for recurrent vomiting. Hyponatremia identified: Serum sodium= 101meq/l. Hypertonic saline (3%) administered.
30/06/22	Serum sodium level – increased to 133 Meq/l.
01/07/22	Started exhibiting behavioral symptoms: confusion, sleep disturbance, increased psychomotor activity. Psychiatry consultation sought. Started on low dose Haloperidol, considering a diagnosis of delirium.
2/07/22	CT Brain taken: enlarged lateral, third, and fourth ventricle. In view of the prominent affective symptoms, a diagnosis of mood disorder considered. Started on Tab. Risperidone.
05/07/22	Discharged from the Department of Medicine. Affective symptoms were persisting at the time of discharge.
14/07/22	Readmitted under Psychiatry in view of the manic presentation, and complaints of unsteadiness of gait.

disturbances, thyroid abnormalities, and CNS infections, including neurosyphilis, was ruled out. There was no history of loss of vision, diplopia, dysphagia, dysarthria, bladder/bowel disturbances, fatigue, or sensory symptoms.

Clinical Findings

There were no signs of malnourishment or liver disease. On examination, there were no signs of limb rigidity or spasticity, reduced power (4+), or meningeal irritation. There were no cerebellar signs, and cranial nerve examination was normal. All deep tendon reflexes were normal, and bilateral plantar was flexor. Tandem gait was impaired.

Her mental status examination at the time of admission to the Psychiatry ward confirmed affective symptoms with psychotic features. She had a cheerful facial expression with increased motor activity and was overfamiliar. Her speech was of increased rate and productivity with increased volume. Thought content revealed increased planning and grandiose ideas. Her affect was elated, and she had lability. She denied any perceptional abnormality. Attention and memory were intact. She had grade 1 insight and impaired personal judgment.

A working diagnosis of Organic manic disorder (according to International Classification of Diseases – Tenth Edition [ICD-10])[7] and Bipolar and related disorders due to cerebral pontine myelinolysis with manic features (according to Diagnostic and Statistical Manual for Mental Disorders – Fifth Edition [DSM-5])[8] was made.

Baseline routine investigations were repeated, were normal, and did not indicate any signs of infection. An MRI of the Brain confirmed both central and extrapontine myelinolysis. Extrapontine involvement was in the caudate nucleus and lentiform nucleus. A CSF study was done to rule out meningitis, and it showed normal results. She was non-reactive for CSF VDRL. CSF CBNAAT was negative, and serum TSH was within normal limits (1.8 mIU/L). She was treated with antipsychotics (Tab Risperidone 4 mg per day) and mood stabilizers (Tab Lithium 800mg per day) and was discharged in two weeks with complete remission of symptoms. Weakness of limbs gradually improved during admission without any specific intervention. The course of event from admission to discharge is mentioned in Table 1.

Consent: Written informed consent was obtained from the patient for this case report.

DISCUSSION

Psychiatric symptoms have not been extensively reported in individuals diagnosed

with central pontine myelinolysis or extrapontine myelinolysis and may be listed as a rare manifestation. If present, they are seen alongside other common neurological motor manifestations. In 2012, in the case report of a 47-year-old male who had an unremarkable medical history, comorbid alcohol use disorder, and worsening of psychotic and bulbar symptoms despite adequate treatment measures, an MRI of the brain showed CPM and EPM.[9] However, in this case report, the influence of substance use disorder in the genesis of symptoms could not be ruled out, especially as the symptoms emerged during the withdrawal period. In the current case report, there was a temporal correlation between the onset of neurological symptoms and mood symptoms.

In keeping with the findings of our study, other authors have reported an association between central pontine myelinolysis and mood symptoms.[10]

A few of the reported neuropsychiatric manifestations include agitated delirium, inappropriate affect, emotional lability, personality changes, poor judgment, emotional incontinence, and disinhibition.[11] Incidentally, some studies have shown increased connectivity between the ventral striatum and thalamus in bipolar disorders.[12] Prevailing literature commonly suggests the involvement of the medial orbital cortex and emotion processing centers involving the hippocampus and amygdala. In our case report, there is involvement of pons and dorsal striatal structures, which may indicate a more complex etiology for affective symptoms and warrant further exploration.

Conclusion

In conclusion, mood symptoms can present as one of the rare manifestations of CPM. We recommend that neuropsychiatric symptoms such as psychotic, mood, or catatonic symptoms developing in the background of rapid correction of low serum sodium be investigated thoroughly and should not be assumed to be a part of a primary psychiatric disorder. Such rare psychiatric presentations of CPM could provide additional insights into the biological correlates in the aetiopathogenesis of specific psychiatric symptoms and, perhaps, mental disorders.

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