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Case Report

Recovery from osmotic demyelination syndrome associated with pituitary macroadenoma

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ABSTRACT

45 years old female known case of hypothyroid admitted to local nursing home with pain abdomen, vomiting and generalized weakness. She received intravenous fluids and antibiotics in a nearby hospital. She experienced hyponatremia, which hypertonic saline from 104 to 128 meq/l quickly treated. Modified Rankin scale score was >3. MRI brain done showed pituitary macroadenoma. She was transferred to our hospital after 5 days because of altered sensorium and bradykinesia of all 4 limbs with high grade fever and unexpected MRI finding. Treatment was started initially in line of meningitis but CSF study was negative. She was intubated for airway protection later tracheostomy was done. MRI brain repeated after 15 days showed pontine and extrapontine demyelination. Serum cortisol was low due to secondary adrenal insufficiency so started on steroid therapy. She improved gradually regained consciousness, started obeying commands, decannulated and discharged with Rankin score <1 after 2 months of steroid and thyronorm supplements and extensive supportive therapy.

1. Introduction

Osmotic demyelination syndrome (ODS) is commonly seen after low sodium correction which was first described by Adams et al. in 1959 in malnourished alcholics who had hyponatremia. Alcoholism and chronic nutritional deficiency were initially considered to be the cause of ODS.[1] Although ODS has long been thought to have a dismal prognosis, some recent reports involving patients who were admitted to clinical wards indicated a less morbid disease with survival and rehabilitation to an independent life.[2] For the sickest ODS patients who need mechanical breathing, very few data are available. The benefit of continuing supportive therapy in the form of costly antibiotics and antifungals for repeated infections because of invasive devices like tracheostomy tube, central line, arterial line and urinary catheters was not guaranteed. We present a case of chronic ODS that, following two months of intensive care management, exhibited full recovery, adding to the ODS cases that recovered after hyponatremia correction.

2. Case Presentation

45 years old female with history of hypothyroidism and dyslipidemia with vomiting for 3 days which was non projectile, four to six episodes per day associated with fatigue and dizziness. On the fourth day of her illness, she was admitted to a local hospital. She was afebrile, with a pulse rate of 104 beats per minute, and a blood pressure reading of 130/80 mmHg when she was admitted to the neighbourhood hospital. Her Glasgow coma scale (GCS) score was 15 out of 15, her pupils were equally responsive to light, and there was no neck stiffness. She was found to have a sodium level of 104 meq/l, which was quickly corrected with hypertonic saline up to 128 meq/l. After

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2 days of sodium adjustment, she experienced decreasing consciousness and decreased verbal response and she had a spike of fever. Modified Rankin scale score was >3. There were no seizures. MRI brain done on 6^{th} day of illness which showed sella-suprasellar mass lesion of size 14mm X 11.4mm X 14mm abutting the optic chiasma. She has no visual symptoms. Analyses of the cerebrospinal fluid (CSF) weren't conducted. On the ninth day of her sickness, she was flown to Kolkata's Apollo Multispecialty Hospital (AMHL) for additional treatment. There was no history of COVID-19 infection, psychiatric disorder and exposure to toxic substances.

Table 1: Biochemical indicators at the nearby hospitalon 4^{th} day

Serum sodium	104meq/l
Serum potassium	2.8meq/l
Serum urea	27.0mg/dl
Serum creatinine	1.3mg/dl

On 5th day serum sodium was 128meq/l

On examination, she has GCS of E2M4V2. She has low grade fever. All four limbs displayed increased muscular tone and hyperreflexia, along with bilateral extensor plantar reflexes. Abdominal, respiratory, and cardiovascular exams were all clear.

Table 2: Hematopathology and biochemistry parameters of the patient at AMHL Haematology

Hemoglobin	11.0g/dl
Total leucocyte count	7000 cells /mm3
Neutrophils	52%
Lymphocytes	40%
Monocytes	04%
Eosinophils	04%
Basophils	00%
Biochemistry	
Glucose (random)	119 mg/dl
Urea	41 mg/dl
Creatinine	1.8 mg/dl
Serum sodium	153meq/l
Serum potassium	3.9meq/l
c-reactive protein	4mg/dl
Aspartate amino transferase	37 U/l
Alanine amino transferase	22 U/l
Alkaline phosphatase	135 U/l
Random cortisol	2.3 mg/dl

She was initially started on ceftriaxone, vancomycin, and acyclovir as CNS infection was suspected. MRI brain done on 9^{th} day of illness showed suprasellar mass lesion of size 14 X 11.4 X 14mm likely suggestive of pituitary macroadenoma. No other significant abnormality seen.

CSF study done on 9^{th} day of illness which showed protein 55 mg/dl, glucose 81mg/dl, Total WBC 20 cells/mm3, lymphocytes 100%, Comprehensive CNS panel and autoimmune panel was negative. Mycobacterium tuberculosis was not detected. Fungal and bacterial cultures were negative. Vancomycin and acyclovir stopped. Screening for sepsis included sputum culture showed klebsiella pneumoniae sensitive to minocycline. Urine and blood cultures and chest x-ray were unremarkable. Sodium level was 153 meq/l on admission to our hospital. Sodium level lowered by D5W and free water gradually.

Serum Cortisol was low due to secondary adrenal insufficiency which was confirmed by ACTH stimulation test, so started on injection hydrocortisone 50 mg intravenous thrice daily. Thyroid stimulating hormone was low though she was on tablet thyronorm 25mcg, increased to 50mcg. Other hormones of pituitary gland like luteinizing hormone, follicle stimulating hormone and growth hormone were within normal limits which indicated a non-secretory pituitary tumor. Her blood sugar levels were within normal limits. She had persistent fever with low GCS E2M3V1 with Rankin score >3, intubated for airway protection. After 5 days on mechanical ventilation, Rankin score was > 3 there was no improvement in sensorium so tracheostomy done and supportive treatment given in neuro intensive care unit. Low molecular weight heparin started for deep venous thrombosis prophylaxis.

MRI brain repeated on 19^{th} day of illness in view of recent severe hyponatremia and correction to rule out osmotic changes in brain which showed altered signal intensity involving bilateral caudate nuclei, bilateral putamina, pons and anterosuperior part of bilateral cerebellar hemispheres including bilateral middle cerebellar peduncles-likely suggestive of pontine and extrapontine myelinolysis.

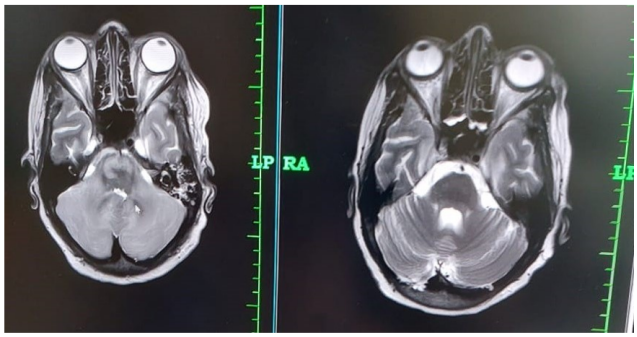

Fig. 1: MRI brain repeated on 19^{th} day of illness

Inj Hydrocortisone was changed to wysolone tablet in a dose of 10mg-5mg-10mg. secondary infections treated with appropriate antibiotics. Ryles tube feeding, Chest and limb physiotherapy given. On 45^{th} day of illness she became drowsy and her sodium level was 114 meq/l corrected slowly with hypertonic saline and her steroid dose was increased wysolone tablet 10mg-10mg-10mg. After nearly 60 days of her illness sensorium improved gradually, Rankin score was <1, obeying commands, mobilized out of

bed, tracheostomy deccanulated and discharged home with steroid and thyronorm supplements.

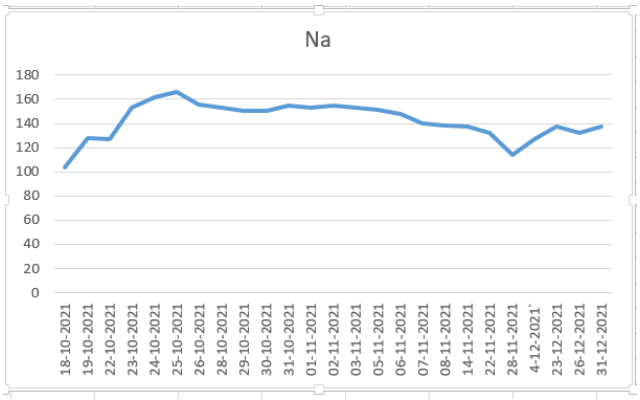

Fig. 2: Variations in blood sodium levels throughout time. The graph indicates a quick correction in sodium levels from a low of 104 to 128 meq/L

3. Discussion

ODS was once believed to have a consistently bad prognosis because the diagnosis was only made by necropsy. However, recent data have demonstrated that complete or nearly complete recovery from ODS is not uncommon, and at the absolute least, remission of severe symptoms is achievable.[1,3] According to a review of the literature, encephalopathy (39%) and hyponatremia (78%) were the two most prevalent predisposing factors to ODS.[1]

ODS symptoms that are currently being experienced include encephalopathy, ataxia, dysarthria, aberrant eye movements, and seizures. 20% of patients had normal first brain MRI results, but after serial imaging, all of them had abnormalities. According to a survey of instances, half of patients experienced a satisfactory recovery while a quarter of patients died. Patients with ODS after liver transplantation had significantly worse outcomes.[4] A good recovery from ODS is not unusual, and a nearly full recovery from severe symptoms is conceivable, according to several recent data.[2–5]

A neurocatastrophe, also known as a catastrophic brain injury, is defined as a trauma severe enough to produce an acute and prolonged loss of consciousness and to significantly increase the risk of death or long-term functional reliance.[6]

If the nonspecific secondary consequences of transient diseases including aspiration pneumonia, ascending urinary tract infection with subsequent septicaemia, deep venous thrombosis, and pulmonary embolism can be avoided, patients with cerebral myelinolysis can live.[5]

Despite initial severe clinical symptoms, the prognosis of critically sick individuals with central or extra pontine myelinolysis is better than it has been so far. Intensivists may underestimate the likelihood of a favourable result given the high rate of decisions to refuse life-sustaining treatments.

4. Conclusions

ODS should be suspected in all patients who had a history of rapid fluctuations in Na even though initial MRI brain was negative. Once ODS was diagnosed extensive supportive therapy like tracheostomy care, chest and limb physiotherapy, ryles tube feeding, bed sore prevention, preventing secondary infections will improve the recovery.

5. Conflicts of Interests

None declared.

Acknowledgement

None.

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