Case Report

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Osmotic demyelination syndrome with transient diabetes insipidus in postpartum female: a case report

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Introduction and importance: Osmotic demyelination syndrome (ODS) is a neurological disorder usually after rapid correction of hyponatremia. Only few cases of ODS with hypernatremia and diabetes insipidus (DI) in postpartum state is reported. Postpartum hypernatremia is described as severe hypernatremia in postpartum period and presents as an encephalopathy with rhabdomyolysis with diffuse white matter hyperintensities suggestive of osmotic demyelination.

Case presentation: The authors present a case of 29-year-old female who presented with chief complaint of altered sensorium and quadriparesis. Two days prior to onset of symptoms, she underwent caesarean section, was kept on nil per oral and free fluid restriction, after which she had confusion, altered sensorium, and weakness in all four limbs. Sodium level was 170 mEq/l. Urine osmolality and plasma osmolality was 150 and 410 mOsm/kg of water, respectively. MRI showed high signal intensity lesion in pons suggestive of demyelination. She was diagnosed ODS with transient DI and quadriparesis, in postpartum period due to further rise in sodium after free fluid restriction and nil per oral. She was treated with desmopressin, 5% dextrose and 0.9% normal saline, her quadriparesis recovered and desmopressin was tapered and stopped over 45 days and discharged at stable state.

Clinical discussion: ODS can rarely be associated with hypernatremia in postpartum female presenting as quadriparesis and altered sensorium.

Conclusion: Clinicians should be familiar of ODS with hypernatremia with transient DI in postpartum period, which is reversible and can be managed by desmopressin and fluid replacement.

Keywords: diabetes insipidus, osmotic demyelination syndrome, postpartum period

Introduction

Osmotic demyelination syndrome (ODS) is a neurological disorder that most frequently arises due to rapid medical correction of hyponatremia[1]. ODS can also be seen in those with a history of malnutrition, alcohol use disorder, chronic liver disease, and hyperemesis gravidarum[2]. Overall, prevalence of ODS ranges from 0.4 to 0.56% among patients with neurological problems visiting tertiary hospital[3]. Majority of studies have shown occurrence of ODS after rapid correction of hyponatremia; however, few studies have been reported for occurrence of ODS in setting of normonatremia or hypernatremia, as in postpartum period or in patients with chronic renal failure who are on dialysis[4].

HIGHLIGHTS

- Osmotic demyelination syndrome can rarely present with hypernatremia in postpartum female.
- This condition is reversible and can be managed by desmopressin and fluid management.

Postpartum hypernatremia is described as severe hypernatremia in postpartum period and presents as an encephalopathy with rhabdomyolysis with diffuse white matter hyperintensities suggestive of osmotic demyelination. The clinical presentation of postpartum hypernatremia includes quadriparesis, delirium, disorientation, irrelevant speech, dysarthria, ataxia, seizures, rhabdomyolysis, hyperuricemia, acute kidney injury, encephalopathy, and coma[5].

Gestational diabetes insipidus (DI), a rare condition that affects some pregnant women, usually in the third trimester. The main cause of this condition is too much vasopressinase, made by placental trophoblasts[6].

The diagnostic modality of choice is MRI scan of head[7]. In the literature, MRI findings of ODS consist primarily of hyperintensity on T2 weighted images or Fluid-attenuated inversion-recovery (FLAIR) images in the central pons (known as central pontine myelinolysis), midbrain, bilateral thalami and/or basal

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ganglia (called as extrapontine myelinolysis), or a combination of these[8].

Here we present a case of 29-year-old female who presented with chief complaint of altered sensorium and quadriparesis, diagnosed as ODS with transient DI after MRI Scan and renal function tests. She underwent caesarean section and was on nil per oral (NPO) and free fluid restriction just 2 days before the onset of symptoms. Then she developed confusion, altered sensorium and weakness in all her four limbs. ODS is usually present with hyponatremia after its rapid correction. However, in this case ODS is associated with postpartum hypernatremia which is rare. The work has been reported in line with CARE guidelines.

Case presentation

A 29-year-old married female, P1L1, housewife by occupation presented with chief complaint of altered sensorium and quadriparesis. Two days before the symptoms began, she underwent caesarean section delivery. She was kept on NPO and was on free fluid restriction for 24 h. Then she had confusion, altered sensorium and weakness in all her four limbs. There was no history of fever, delusion, nausea, vomiting, seizures, weakness of the face, problems in swallowing, or slurred speech. She did not give a history of any past illness. She consumes a non-veg diet, does not smoke or consume alcohol, and had regular menstruation before her pregnancy. There is no family history of a similar illness.

On examination her general condition was poor (GCS 7/15). She was not oriented to time, place and person. Her vital signs were stable and within normal limits. There was no pallor, icterus, lymphadenopathy, oedema, cyanosis, or clubbing. There was a weakness of all upper and lower extremities with no facial involvement (muscle power was 3/5). The rest of the systemic examination findings were regular.

Investigations such as liver function tests, renal function tests, haematologic tests, urine and plasma osmolality, MRI scan, hormone profile tests were done as shown in Table 1. MRI scan showed multiple T2/FLAIR high signal intensity lesion in the pons suggestive of central pontine demyelination as shown in Figs. 1 and 2. Sodium level was 170 mEq/l. Urine osmolality and plasma osmolality was 150 and 410 mOsm/kg, respectively, with high urine output, suggestive of DI. She was diagnosed as ODS with DI and quadriparesis, in postpartum period due to rise in sodium after free fluid restriction and NPO.N

She received desmopressin, 5% dextrose and 0.9% normal saline as treatment, which helped her regain her muscle strength in all four limbs. She gradually reduced and stopped taking desmopressin over 45 days and left the hospital in a stable condition.

Table 1

Investigations.

Tests	Units	Results	Reference range
Sodium	mEq/l	170	135–146
Potassium	mEq/l	4.2	3.5–5.2
Urea	mmol/l	12.7	2.8–7.2
Creatinine	mmol/l	77	45–84
Plasma osmolality	mOsm/kg	410	mOsm/kg
Urine osmolality	mOsm/kg	150	mOsm/kg
TLC	/cmm	7600	4000–11 000
Hb	gm%	14.9	12.5–15.0
Platelets	/cumm	656000	150 000–400 000

Hb, haemoglobin; TLC, total leucocyte count.

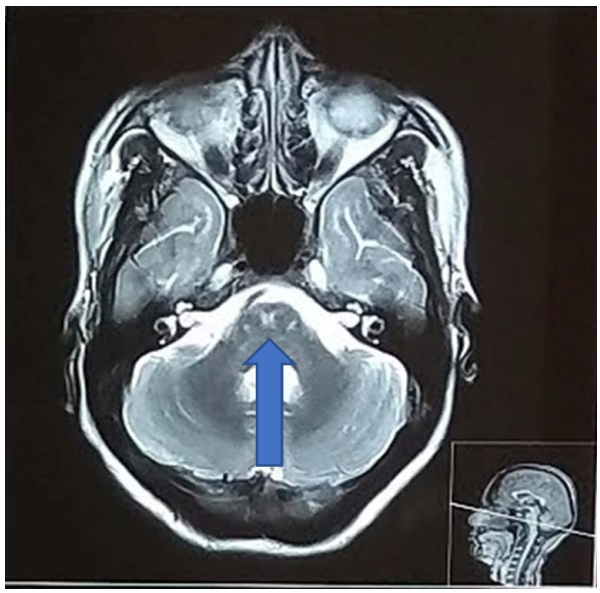

Figure 1. T2-weighted image showing high signal intensity lesion in pons.

Discussion

ODS is usually caused by rapid correction of hyponatremia, rarely it might present with normonatremia or acute hypernatremia. Many theories have revealed the pathophysiology behind this, is the injury to grey matter rich regions by myelino-toxic substances released as a result of osmotic injury to endothelium[4]. ODS can present as central pontine myelinolysis involving white matter tract of pons or extrapontine myelinolysis

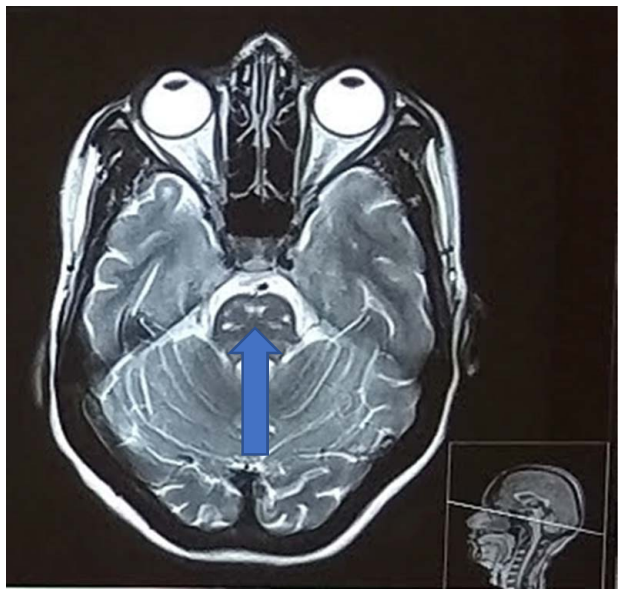

Figure 2. T2-weighted image showing high signal intensity lesion in pons.

involving extrapontine areas 9. Hypernatremia usually presents with various neurological manifestations from flaccid paralysis to impaired cognition, encephalopathy, and deep coma[3]. Most report and studies suggest that ODS happens when hyponatremia is corrected too fast. However, a few reports show that it can happen with normal or high sodium levels, such as in postpartum patients[4]. Here, we present a case of ODS caused by hypernatremia due to free fluid restriction in postpartum female.

The clinical manifestations of ODS can involve various regions of the brain depending upon location and severity of demyelination. The symptom may include altered mental status, dysarthria, dysphagia, pseudobulbar symptoms, quadriparesis, and extrapyramidal symptoms[9]. In our case pons is involved and presented with confusion, altered sensorium, and quadriparesis.

The diagnosis of ODS is usually confirmed on the basis of clinical presentation and radiological findings. MRI is the most sensitive imaging modality for detection of characteristic lesions in the brain[10]. In the central pontine myelinolysis, MRI findings include hyperintensity on T2/FLAIR images in the pons and similar findings involving the basal ganglia, thalamus, lateral geniculate body, and cerebellum is present in MRI in case of extrapontine myelinolysis[11]. Here in our case MRI scan revealed multiple high signal intensity lesion in pons suggestive of central pontine myelinolysis.

There is no proven treatment for ODS apart from supportive care. However, lowering of sodium level with the help of 5% dextrose and desmopressin have shown beneficial effect in treating ODS in animal models but data in humans is limited to case report and case series[12]. In this case, our patient was treated with 5% dextrose, desmopressin, and 0.9% saline. Her condition was improved and she regained her muscle power. Desmopressin was gradually tapered and she was in stable state during discharge.

Conclusion

Clinicians should be aware about the unusual presentation of ODS associated with transient DI in postpartum female which can present with altered sensorium and quadriparesis. This condition is reversible and can be managed by 5% dextrose and desmopressin.

Author agreement statement

We the undersigned declare that this manuscript is original, has not been published before and is not currently being considered for publication elsewhere. We confirm that the manuscript has been read and approved by all named authors and that there are no other persons who satisfied the criteria for authorship but are not listed. We further confirm that the order of authors listed in the manuscript has been approved by all of us. We understand that the Corresponding Author is the sole contact for the Editorial process. He/she is responsible for communicating with the other authors about progress, submissions of revisions and final approval of proofs.

Ethical approval

This is a case report. Therefore, it did not require ethical approval from the ethics committee.

Consent

Written informed consent was obtained from the patient for publication of this case report. A copy of the written consent is available for review by the editor-in-chief of this journal on request.

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Author contribution

N.P.: involved in counselling and treatment of the patient. A.Y., A.S., B.B., K.K.K.: collected all the required information, reports, figures; reviewed the literature and contributed in writing and editing the manuscript. All authors read and approved the final manuscript.

Conflicts of interest disclosure

None.

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