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General Medicine

RARE PRESENTATION OF SHEEHAN'S SYNDROME AT A TERTIARY CARE CENTRE: A CASE REPORT

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ABSTRACT Sheehan's syndrome is a rare fatal complication that occurs due to ischemic necrosis of the pituitary gland. It was identified in 1937 by Sheehan. It is characterized by anterior pituitary dysfunction due to postpartum necrosis of the pituitary gland after massive bleeding. Our patient presented to the institution during September 2022 with history of severe vomiting for 2 days and altered sensorium for 1 day. She had hyperpigmentation on the face. She is lethargic and not oriented to time, place and person. Pupils were normal in size and reactive to light. There were no signs of meningeal irritation and there is no skull or spine tenderness. She was found to have hyponatremia as per serum electrolytes test. She was given 3% NACL after admission for correcting hyponatremia. Chest x ray, CT chest and abdomen were normal. In view of history of amenorrhea since last 12 to 14 years and lactational failure after last child birth (Hypogonadism) and inappropriately normal TSH for low T4 (Central Hypothyroidism) and inadequate cortisol release to ACTH-stimulation test, secondary adrenal insufficiency due to panhypopituitarism is suspected. So, MRI of brain was done which showed empty sella with thinned out pituitary. She was kept on Intravenous hydrocortisone and was discharged in stable condition 1 week after admission with a prescription of oral prednisolone, levothyroxine, oral calcium, B-complex and vitamin D3 supplementation.

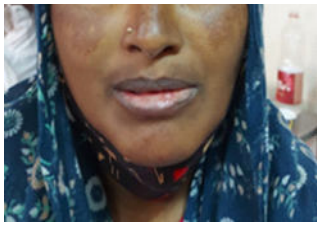
KEYWORDS : Altered mental sensorium, Hormonal profile, Hypopituitarism, Hypothyroidism, Sheehan's syndrome

Introduction

Sheehan's syndrome is a rare fatal complication that occurs due to ischemic necrosis of the pituitary gland.1 It was identified in 1937 by Sheehan.2 It is characterized by anterior pituitary dysfunction due to postpartum necrosis of the pituitary gland after massive bleeding. It is a rare complication that happens in around 5 among 100,0003-4 births globally and is the commonest reason of hypopituitarism in developing countries, which could be due to lack of proper access to advanced medical procedures and skilled professionals. The syndrome constitutes for around 0.5% of all known cases of hypopituitarism among females.5 The prevalence is very high in India and it was 3.1% in a state, where more than 50% of affected patients had home deliveries.6 Infarction of enlarged anterior pituitary lob's size due to hyperplasia of cells that secrete prolactin due to increased estrogen section is the pathogenesis behind Sheehan's syndrome. It also occurs due to compression of blood vessels supplying pituitary gland by enlarged gland or due to significantly reduced blood supply during intrapartum or postpartum period. Other reasons include autoimmunity, vasospasm, small sella, disseminated intravascular coagulation (DIC).7 It can present during several months or years after delivery. One study done in France showed that there is a delay of 9.7 years in diagnosing Sheehan's syndrome, and a delay of 20 years was also seen in developing countries.8 Females with Sheehan's syndrome can show panhypopituitarism or just selective pituitary deficiencies.9-10 One of the common symptoms of Sheehan's syndrome include amenorrhea. Sometimes, it can present as an emergency condition with severe hyponatremia, circulatory collapse, hypoglycemia, diabetes insipidus, cardiac failure, or psychosis.11 Sometimes, the diagnosis can't be made unless patient show features of secondary hypothyroidism or secondary adrenal insufficiency.12 Anemia can be seen commonly. Pancytopenia can also be seen rarely. We present a case of Sheehan's syndrome in a 40-year-old female. Informed consent was taken from the patient before publishing this case and taking photographs of her and using her data.

Case Presentation: A 40-year-old housewife, presented to our institution on September 2022 with a history of severe vomiting for 2 days and altered sensorium for 1 day. She had recurrent episodes of vomiting 3-4 times a day, which is non-bilious, non-projectile, with ingested food/fluid as its content. Mental sensorium was altered suddenly since 1day, and the patient was unable to recognise her own family members. There is a decreased responsiveness to verbal commands. Relatives gave a history of easy fatiguability since last 1 week. There is no history of fever, cough, cold, sore throat, abdominal pain, burning micturition, passage of loose stools, headache, blurring of vision, double vision, projectile vomiting, trauma to head, loss of consciousness, seizures, jaundice, rash over body, petechiae/purpura, decreased urine output, pedal edema, facial puffiness, drug/medication intake/toxin exposure, recent vaccination, dog bite, recent travel history. But she had similar complaints 4 years back, for which she was taken to a private hospital and was given intravenous sodium and then, she was recovered. She had no history of diabetes, hypertension, coronary artery disease, kidney disease, lung disease, asthma, hypothyroidism, pulmonary tuberculosis, obstructive pulmonary disease, allergies. There is no history of malignancy in the family. Patient takes mixed diet, sleep was adequate and her appetite was normal. Bowel and bladder habits were regular. She has no addictions. Differential diagnosis includes metabolic or toxic or vascular or infective or inflammatory or neoplastic causes. On general examination, patient is drowsy with no orientation to time, place, person. Glasgow coma scale shows E4V1M3 stage. She is moderately built and nourished. Her BMI is normal. There is no pallor, cyanosis, icterus, clubbing, lymphadenopathy, and pedal edema. She had hyperpigmentation on face.

Figure 1 shows hyperpigmentation of face



Vitals showed normal pulse rate, normal blood pressure and normal respiratory rate. Her random blood glucose was 108mg/dl. Oxygen saturation was 98% on room air. Systemic examination showed normal S1 and S2 sounds, non-tender, soft abdomen without any organomegaly. Pupils were normal in size and reactive to light. There were no signs of meningeal irritation and there is no skull or spine tenderness.

Table 1 shows her serum sodium (Na) and potassium(K) levels each day, which showed gradual increase in sodium levels.

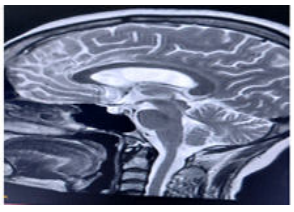
Date	Day 0	Day 1	Day 2	Day 3
S.Na	118	117	121	123
S.K	3.7	4.0	3.8	4.1

Patient was given 3% NACL after admission.

On day 2, Urine osmolality was 508 mOsm/kg. spot urine Na+ was 123mEq/L, spot urine creatinine was 55mg/dL, spot urine chloride was 118 mEq/L and urine for K+ was 25.28mEq/L. Hence a

provisional diagnosis of hypotonic hyponatremia was made. Patient was euvolemic and hence the possibility of syndrome of inappropriate antidiuretic hormone (SIADH) was considered. Chest x ray, CT chest and abdomen were normal. TSH levels were inappropriately normal. Serum 8 AM Cortisol was 2.67 mcg/dl which was very low and ssuggestive of adrenal insufficiency. ACTH stimulation test was done using Acton Prolongatum and post-stimulation cortisol levels were 14.7 mcg/dL, indicating inadequate response, conforming adrenal insufficiency. On further examination, patient has no mucosal hyperpigmentation or goitre but she had amenorrhea since last 14 years and lactation failure after the birth of 3rd child. In view of history of amenorrhea since last 12 years and lactational failure after last child birth (hypogonadism) and inappropriately normal TSH for low T4 (central hypothyroidism) and inadequate cortisol release to ACTH-stimulation test, secondary adrenal insufficiency due to panhypopituitarism is suspected. So, MRI of brain was done which showed empty sella with thinned out pituitary.

Figure 2 showing empty sella with thinned out pituatary gland in MRI brain



Follicle stimulating hormone, luteinizing hormone, prolactin levels were decreased. The patient is diagnosed to have SHEEHAN'S SYNDROME with empty sella. Treatment was started with Injection Hydrocortisone 100mg 8th hourly. After starting the treatment, S. Sodium levels increased slowly and treatment was shifted to oral hydrocortisone three times a day in dose of 10, 10 and 5 mg. After 2 days of hydrocortisone supplementation, thyroxine supplementation was started at a dose of 50mcg/day. As patient developed hypertension and hyperkalaemia, oral prednisolone was given instead of hydrocortisone. Patient became normotensive and serum sodium, potassium levels returned to normal. Patient was discharged in stable condition 1 week after admission with a prescription of oral prednisolone, levothyroxine, oral calcium, B-complex and vitamin D3 supplementation.

Discussion:

Diagnosis of Sheehan's syndrome is determined by patient's history, physical examination, and hormonal tests. Lab tests may reveal hyponatremia, which is most common electrolyte imbalance, that occurs in 33% to 69% of patients.14-15 Anemia is a common and recognized feature of hypopituitarism. We reported a case of recurrent severe symptomatic hyponatremia, which is defined as serum sodium ≤120 mEq/L. The patient had hypotonic hyponatremia with high urine osmolality and high urine sodium excretion, which represented the effect of ADH and euvolemic status. Very low serum uric acid and blood urea nitrogen in this euvolemic hyponatremia patient suggested SIADH, which is the most common cause of euvolemic hyponatremia. Continuous secretion of ADH despite hypo-osmolality and normal or increased effective circulatory volume results in impaired water excretion and hyponatremia. Various medical conditions can cause SIADH, which includes malignancy, pulmonary diseases, central nervous system disorders, and various medications. Although most patients who were diagnosed with SIADH had no identifiable etiology, SIADH might be the first presentation of the primary occult disorder. Our patient did not take any medication and had normal chest radiography. Serum morning cortisol was low. Further investigations, including ACTH level and ACTH stimulation test, were compatible with secondary adrenal insufficiency. Severe hyponatremia was dramatically improved after glucocorticoid replacement, which suggested the aetiologies of hyponatremia from adrenal insufficiency. Hormonal assays and MRI of pituitary gland revealed hypopituitarism due to empty sella secondary to Sheehan's Syndrome. An appropriate investigation for hyponatremia and attentive workup for secondary causes of SIADH led to a diagnosis of hypopituitarism from empty sella syndrome and precise treatment with hormone replacement therapy in this patient.

Fatma M reported Sheehan's syndrome in a 48-year-old female who presented with vomiting, confusion, abdominal pain and diarrhea and the patient had pancytopenia.13 Gokalp reported hematological abnormalities in patients with Sheehan's syndrome. The prevalence of anemia was 80% compared to 25% among controls.16 Hypothyroidism, hormonal deficiency, adrenal insufficiency may explain normochromic anemia in hypopituitarism.17 It can also occur due to physiologic adjustment to lower oxygen requirement, as pituitary hormones modify erythropoietin synthesis in the kidney.[18]

Conclusion: Our case report highlights the importance of proper investigation of underlying causes of hyponatremia apart from correction of sodium levels in patients with hyponatremia.

There were no conflicts of interest.

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